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Long-term results after single event multilevel surgery for the correction of gait disorders in spastic diplegic cerebral palsy

PhD dissertation

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Abbreviations

2D: two dimensional

3D: three dimensional

A/D converter: analog digital converter

ANOVA: analysis of variance

APT: anterior pelvic tilt

BMI: body mass index

BTX: botulinum toxin type A

CBM: conversion of biarticular to monoarticular muscles

CC-Distraction: calcaneo-cuboid-joint distraction arthrodesis

C-DRFT: group that had distal rectus femoris transfer to correct decreased peak knee flexion in swing phase

CHL: combined medial and lateral hamstring lengthening

CNS: central nervous system

CP: cerebral palsy

DF: dorsiflexion

DRFT: distal rectus femoris transfer

E0: preoperative examination

E1: examination 1 year postoperatively

E2: second examination after surgery

E3: third examination after surgery

EMG: electromyography

FDO: femoral derotation osteotomy

GC: gait cycle

GGI: Gillette Gait Index

GMFCS: Gross Motor Function Classification Scale

GRF: ground reaction force

MHL: medial hamstring lengthening

MRC: medical research council

MTL: muscle tendon lengthening

PASW: public art south west

P-DRFT: group that had prophylactic distal rectus femoris transfer

PF: plantar flexion

pKFSw: peak knee flexion in swing

ROM: range of motion

SD: standard deviation

SEMLS: single event multilevel surgery

SENIAM: surface EMG for a non-invasive assessment of muscles

SPSS: statistics statistical procedures companion

TDO: tibial derotation osteotomy

UK: United Kingdom

USA: United States of America

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1. Introduction

1.1. Definition

Cerebral palsy (CP) is a childhood condition in which there is a motor disability (palsy) caused by a static, non-progressive lesion in the brain (cerebral). The causative event has to occur in early childhood usually defined as less than 2 years of age. Children with CP have a condition that is stable and non-progressive; therefore, they are in most ways normal children with special needs (1).

The prevalence of cerebral palsy is 2-3 children from 1000 neonate (2). The most commonly reasons are premature and low birth weight (<1500g).

1.2. Etiology

Etiological factors can be separated into a time period as to when these insults occurred: congenital, neonatal and postnatal. (1. Table, (3)) Congenital deformities result from defects that occur in normal development and follow patterns based on failures of normal formation. In neonatal deformities the newborn suffers some injury during the birth. Postnatal causes may overlap somewhat with the prenatal and neonatal group. Most of infections are prenatal and neonatal viral infections (CMV, VZV), but postnatal deformities can be caused by bacterial meningitis. Children, who survive this infection have a 30-50% chance to become CP.

1. Table: Etiology of cerebral palsy

Congenital	**Neonatal**	**Postnatal**
defect in the development of neural tube or brain infection alcohol, nicotine	hypoxic events cerebral hemorrhages neonatal stroke Kernicterus	postnatal trauma (shaken baby syndrome, blunt head trauma) metabolic encephalopathy infections toxicins

A specific region of brain injury can cause variation in the impairments because the initial injury also hinders normal development. Because all these injuries occur in the young and immature brain, growth and development over time affects the impairment. During healthy development, the early primitive reflexes (Moro reflex, sucking reflex, tonic labyrinth reflex, asymmetric tonic neck reflex, step reflex, parachute reaction etc.) should disappear. The preservation of these could indicate the presence of brain injury.

1.3. Pathology

Cerebral palsy requires a pathologic lesion in the brain, in the central motor system (3). In most cases the spinal cord has a normal function, but there are some children who have a primary lesion there. The control of motion is either volitional or automatic. The automatic response is a relatively simple neuronal reflex at the spinal cord level. All volitional motion initiates in the cerebral cortex and transmitted to the peripheral motor nerves through the cortical spinal tracts traversing the internal capsule and the spinal cord.

The peripheral motor system includes the nerves and the musculoskeletal system. In CP children there are no primary lesion in any of peripheral systems. The impairment of the central motor system causes secondary abnormal development on the periphery.

Patterns of CP can be categorized further by clinical presence quality or by topography of palsy (4). The quality can be spasticity, athetosis, dystonia and rigid quality of palsy. Spasticity is the most frequent form (more than 80%). It is caused by co-contraction by agonist and antagonist muscle. This is a velocity dependent increase in resistance to motion. Athetosis (6%) is large, involuntary, slow cramped motions of the more proximal joints, which can be exacerbated by mental strain. Dystonia is a slow motion with a torsional element, which may be localized to one limb or involve the whole body (9%). The appearance of rigidity is under 5%.

Balance is the maintenance of posture, when impaired children overcompensate for a movement and unable to stand in one place. Ataxia (15%) is the term used to mean abnormal balance.

Topographically CP can be hemiplegic, diplegic and quadriplegic. Hemiplegic involves one half of the body. The flexion is typical in upper extremity and the extension in lower extremity. These children can usually learn to walk. Diplegia involves primarly the lower extremity with extension and have a mild upper extremity involvement with flexion. They require frequently orthosis and other aids to walk. The quadriplegic patients are usually mentally retarded (75%) and have highly spasticity in all four limbs. Only 10% can learn to walk.

Normal muscle tone is composed of stretch reflex, central control of stretch reflex and reflex activity of spine cord. The opposite of spasticity is hypotonia, which means decreased muscle tension when the joint is moved. Spasticity includes an increased sensitivity of the normal stretch reflex in addition to a velocity-dependent increase in resistance, which initiates a muscle contraction to resist the motion (5). To measure the muscle tone most commonly the Ashworth scale or the modified Ashworth scale is used, however it is a very subjective measurement (2. Table) (6, 7).

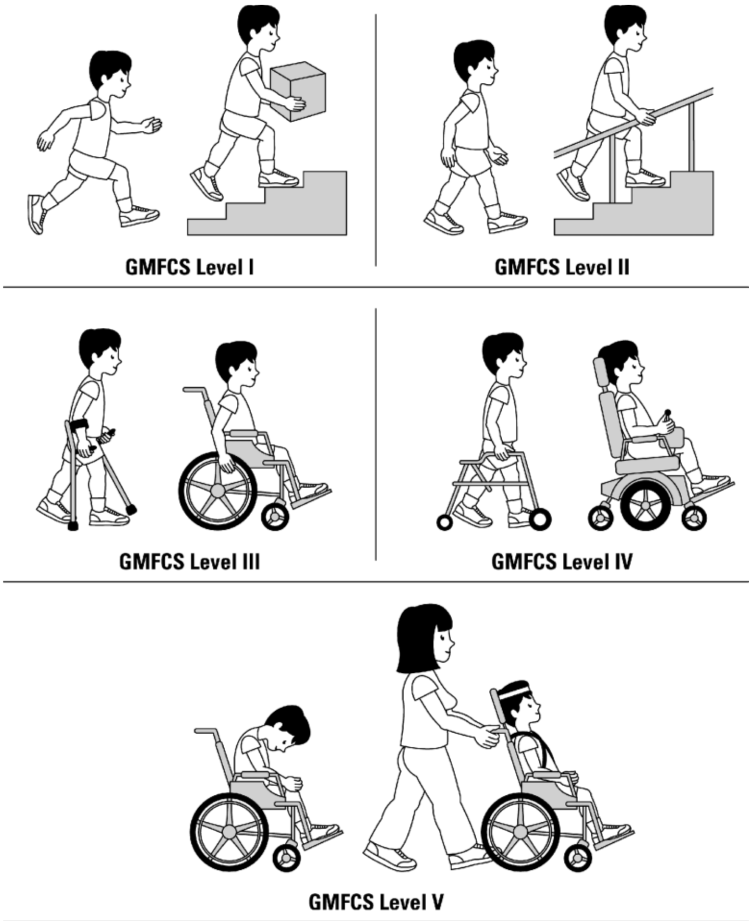
2. Table: Modified Ashworth scale (6)

0	normale tone
1	slight increase in tone, a catch and release at the end of the range of motion (ROM)
1,5	slight increase in tone, a catch followed by minimal resistance in remainder of range
2	more marked increase in muscle tone through most of the ROM, but affected part(s) easily moved
3	considerable increase in tone, passive movement difficult
4	affected parts rigid in flexion or extension

The effects of spasticity on the skeletal muscle include shorter fiber length, decreased volume of the muscle, change in the fiber type and neuromotor junction type. These changes result in weaker muscle and decreased joint range of motion (8). The muscle and tendon growth can not follow the bone growth resulting motion constriction. In the long term, spasticity cause muscle shortening in agonist and muscle elongation in antagonist, resulting in malposition. Especially the multijoint muscles tend to spastic shortening (9, 10). The untreated deformity becomes more severe with growing. First it is still able to change dynamically, but it can be fixed with time resulting in constrictions. These constrictions are followed by the bony malformations. The efficiency of therapy diminishes with growing, but it is hard to say the appropriate time for it. Some surgeons prefer to wait until patients are 8-10 years and perform all of their surgical interventions in one sitting. According to Sussman and Aiona different deformities should be operated in different time points (11).

1.4. Gross Motor Function Classification Scale (GMFCS)

The Gross Motor Function Classification Scale (GMFCS) is a simple five-level system to describe gross motor function in cerebral palsy patients (1.Fig.). The GMFCS I children walk indoors and outdoors and climb stairs without any limitation. Children perform gross motor skills including running and jumping, but speed, balance and coordination are impaired. The GMFCS II children are limited in walking on uneven surfaces and inclines and walking in crowds or confined spaces. They have only minimal ability to run and jump. GMFCS III children can walk only with an assistive mobility device. They may climb stairs holding onto a railing. To take a long distance these children may need a wheelchair. GMFCS IV children may walk for a short distance on a walker. They may achieve self-mobility using a power wheelchair. GMFCS V children are unable for independent mobility. All areas of motor function are limited (12, 13).



1. Figure: Gross motor function classification scale (13)

1.5. Diagnosis

The maturation of neonates take place from infancy to one year of age rapidly from proximal to distal. Children have first head control, then develop the ability to bear weight on the arms, followed by the trunk control, then develop the ability to stand. There are no agreed-upon diagnostic criteria to make a diagnosis of CP. When a child is not meeting developmental milestones, has persistent primitive reflexes, or has significant abnormalities in the elements of motor function, a diagnosis of CP can be made. The history should clearly demonstrate that this is a nonprogressive and is nonfamilial (3). The lack of balance and abnormalities in motor tone are the most common abnormalities that occur in children with CP. But there is a well-recognized phenomenon of children occasionally outgrowing CP. For this reason, it is preferred to make the diagnosis in young children only when it is clear and without doubt, but wait until least age 2 years for children who have more mild and questionable signs.

1.6. Therapy

Most of the CP children receive therapy and go to the school (14). Therapy will be ordered by physicians and medical doctors. It begins already in infancy. Early therapy is provided in a medically-based construct. As the children get older, the main intervention shifts to the educational system. In addition to the standard therapy treatment (physical, occupational, speech therapy, orthotics, pharmacological therapy, surgery), there are many treatment modalities that are promoted as beneficial for CP treatment: hippotherapy, hyperbaric oxygen therapy, hydrotherapy, acupuncture etc. The goal of different therapeutic methods is to decrease the increased muscle tone and to preserve the length of the muscles. The first aim is to prevent those provocative factors that give rise to the problems. The second aim is to prevent the consequences of spasticity (15).

1.6.1. Physical, occupational and speech therapy

Physical therapy focuses on gross motor function, such as walking, running, jumping and joint range of motion (14). It prevents some spasticity from developing and worsening or complications from arising. Its first aim is the muscle lengthening (15). In occupational therapy the main focus is on fine motor skills, specifically upper extremity function and activities of daily living such as dressing, toileting and bathing. The focus depends on the age and functional ability of a child. The therapist uses a learning approach based on specific task as the goal. Speech therapy focuses on oral motor activities such as speech, chewing and swallowing. A major focus of all therapy is to maximize the individual's independence.

1.6.2. Medical equipment

Durable medical equipment is the category of devices that are prescribed to ameliorate the disabilities from the motor impairments (16). Each of devices has a very specific indications and contraindications. The medical equipment has a very wide range of application. It assists a limb positioning with orthosis or sitting with seating system. To prevent a contracture the patient needs daily physical therapy and stretching the joints with orthosis. Most children with CP, at some time during their growth and development, use a walking device. It makes the parents and caretakers very happy, if their child became ambulatory with a walking aids. To assist walking there are many devices: forward- based walker, posterior walker, crutches etc.

1.6.3. Orthosis

Orthosis is externally used medical equipment, which supports the structure and function of neuromuscular system. Its goal is to restore the functions of the joints (17). The orthosis is named for the joints that are crossed by the orthotic (16). The basic aim is always to influence the muscle length and muscle tone. In the literature, a minimum time of 6 hours daily is recommended (18). The more influence the pathological forces have, the more beneficial the orthosis treatment applies. Each orthosis maintenance can be assisted by other conservative measures. An orthosis can only be effective if the related deformity at least partially still can be corrected passively. There are three different orthosis types: orthosis for upper extremities, for spine and for under extremities. Orthosis have different types depending on the anatomical location they applied, for example in the case of the ankle: solid ankle-foot orthosis (AFO), ground reaction AFO, articulated ground reaction AFO, half-height AFO.

While in the past the emphasis was put on the elimination of the consequences of spasticity of the musculoskeletal system, nowadays the modulation of spasticity itself became an important role. The improvement of function and enhancement of the quality of life are also available for this therapeutic approach (19).

1.6.4. Drugs

Pharmacological treatments are available for spasticity of both patterns (15). Oral agents can be used for generalized spasticity and botulinum toxin or phenol injections for focal spasticity. There is no evidence to suggest the best time to introduce pharmacological treatments, but there is evidence for the effectiveness of a variety of oral agents. These include baclofen, dantrolene, tizanidine and benzodiazepines. When physical therapy does not afford sufficient control, supplementary intervention is necessary.

Intrathecal baclofen is frequently mentioned in relation to the treatment of spasticity. However, the numbers of patients who require this therapy will always be small, because of the costs of it. It is designed particularly for people who cannot tolerate oral antispastic agents and is extremely effective in reducing the tone and disability due to lower limb spasticity.

Nerve blockade with phenol or alcohol are also used to treat regional spasticity. Phenol causes fewer problems than alcohol and is a useful adjunct to other forms of focal spasticity treatment, like botulinum toxin A. It is cheap and very effective, but the procedure itself is time consuming.

When focal spasticity is considered, botulinum toxin A is the most widely used treatment. It is indicated where the spasticity is harming the spasticity. It is not used to treat contracture, which is treated using physical means alone. In determining when to intervene, this drug should be given when the spasticity causes functional problem. Some clinicians are also finding that this treatment can be given as a prophylactic agent in certain patients soon after brain injury. There is evidence that the outcomes of physiotherapy improve when used in conjunction with botulinum toxin A treatment.

1.6.5. Surgery

In cerebral palsy the primary disorders of the central nervous system (CNS) can hardly be influenced by surgery (20). Spasticity itself can only be influenced secondarily by reducing the stretch reflex after muscle and tendon lengthening (6). The secondary disorders display the primary target of surgical treatment. Tertiary problems, which represent compensatory mechanisms, normally should vanish after correction of secondary problems and therefore need rarely to be treated. Only if tertiary problems become fixed treatment may be needed. Based on the indications the surgical interventions can be grouped: prophylactic indication, therapeutic indication and palliative indication. Surgical treatment can be done on soft tissue, bones, joints or on central or peripheral nervous system.

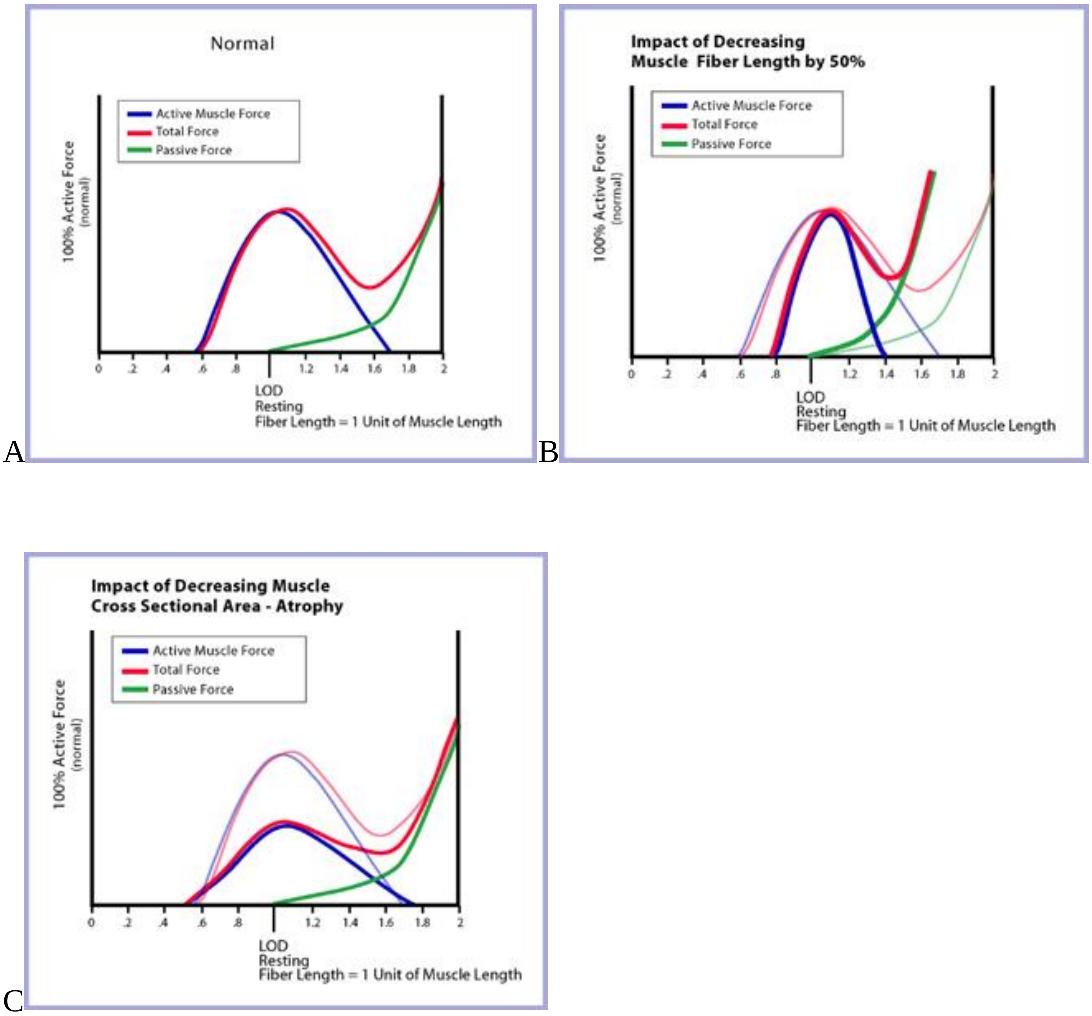
Operations on soft tissue are the myotomy, muscle tendon lengthening, tenotomy, tenodese, tendon transfer, capsulotomy and capsulodesis. The definition of myotomy is a partial or a full ablation in the near of the muscle origin. This technique will be done if a tendon or intramuscular lengthening cannot be allowed. It is typical on the legs by the head of gastrocnemius (operation after Silverskjöld). Tendon lengthening is an incision on the tendon intramuscular or Z-form lengthening and suture on the tendon. It can be done on one or multilevel. On the lower extremity most of the muscle can lengthened: psoas muscle, ischiocrurale muscle, hamstrings etc. It is called tenotomy if the whole tendon will be cut. Typical indication field is on the proximal rectus muscle or on iliopsoas muscle. If a tendon will be sectioned at the end of muscle and over a joint it will be fixed on the bone, it is called tenodese. After this operation the joint will be blocked. Tendon transfer is more common used, then a tenodese. The tendon will be transferred on another muscle to improve the effect of the muscle. The distal end of rectus muscle is often transferred to gracilis or semitendinosus muscle. By cutting the joint capsule to correct the malformation of a joint is called capsulotomy. Under capsulodesis the displacement and a new fixation of a joint capsule in a corrected place is meant. To correct fixed contractures, bony mal-alignment or joint instability, osteotomies or arthrodesis may be needed The transection of the bone and fixation in desired position called osteotomy.

1.6.5.1. Single event multilevel surgery (SEMLS), muscle tendon lengthening (MTL)

Patients with diplegic cerebral palsy show a variety of gait disorders, of which crouch knee gait is one of the most frequent (21-23). Abnormal muscle tone is accused to be a primary factor for gait disorders in the presence of cerebral palsy (24-27). These gait problems are commonly treated by single event multilevel surgery (SEMLS) (26, 28-30). SEMLS refers to the correction of all orthopedic deformities in one session and can be defined as at least two orthopedic procedures at different anatomical sites in each limb (i.e. a minimum of four procedures) (29). The bony and soft tissue operations are combined to adjust joints (hip, knee, ankle) in one session. Various operative principles are available to correct the deformities (lengthening of tendons, transfer of tendons, muscle lengthening, derotation osteotomy, extension osteotomy, stabilization) (6, 31-35).

A component of SEMLS is the MTL, which is commonly used to treat flexed knee gait. It can be performed on intramuscular, aponeurotic and tendon. MTL does not directly treat spasticity but only addresses the secondary effects of decreased muscle growth.

The Blix curve, which represents the muscle fiber length-tension relation, consist of an active and a passive part (36). The muscle excursion corresponds to the resting muscle length, which is achieved by passive stretch. At resting length the muscle has the ability to generate the highest amount of active force. As the muscle shortens, this ability to generate force decreases to zero at approximately 60% of rest length. As the muscle lengthens, the active force-generating ability also decreases and reaches zero at approximately 170% of rest length. However, as the muscle lengthens, the passive collagen elements provide a passive restraint to further lengthening, thereby increasing tension as the muscle is lengthened. This increases until approximately 200% of resting length, when the muscle starts to physically fail (Figure 2A). Muscle shortening seen in children with spastic CP leads to the frequently observed decreased joint range of motion. This shortening of the muscle fiber also leads to significant changes in the length-tension response of the muscle. The effect of decreased muscle fiber length causes a great narrowing of the length-tension curve, meaning that the muscle can generate effective force over a much shorter range. This change concentrates the muscle force-generating ability into a very narrow range of joint (Figure 2B). In addition, many children have decreased muscle diameters, causing muscle weakness defined as having a decreased ability to generate maximum force. This atrophy or weakness causes the peak tension of the length-tension curve to be decreased (Figure 2C).



2. Figure: Blix curve. Fig. 2A: Normal, Fig. 2B: Impact of decreasing muscle fiber length by 50%; Fig. 2C: Atrophy (37)

1.6.5.1.1. Hamstring lengthening

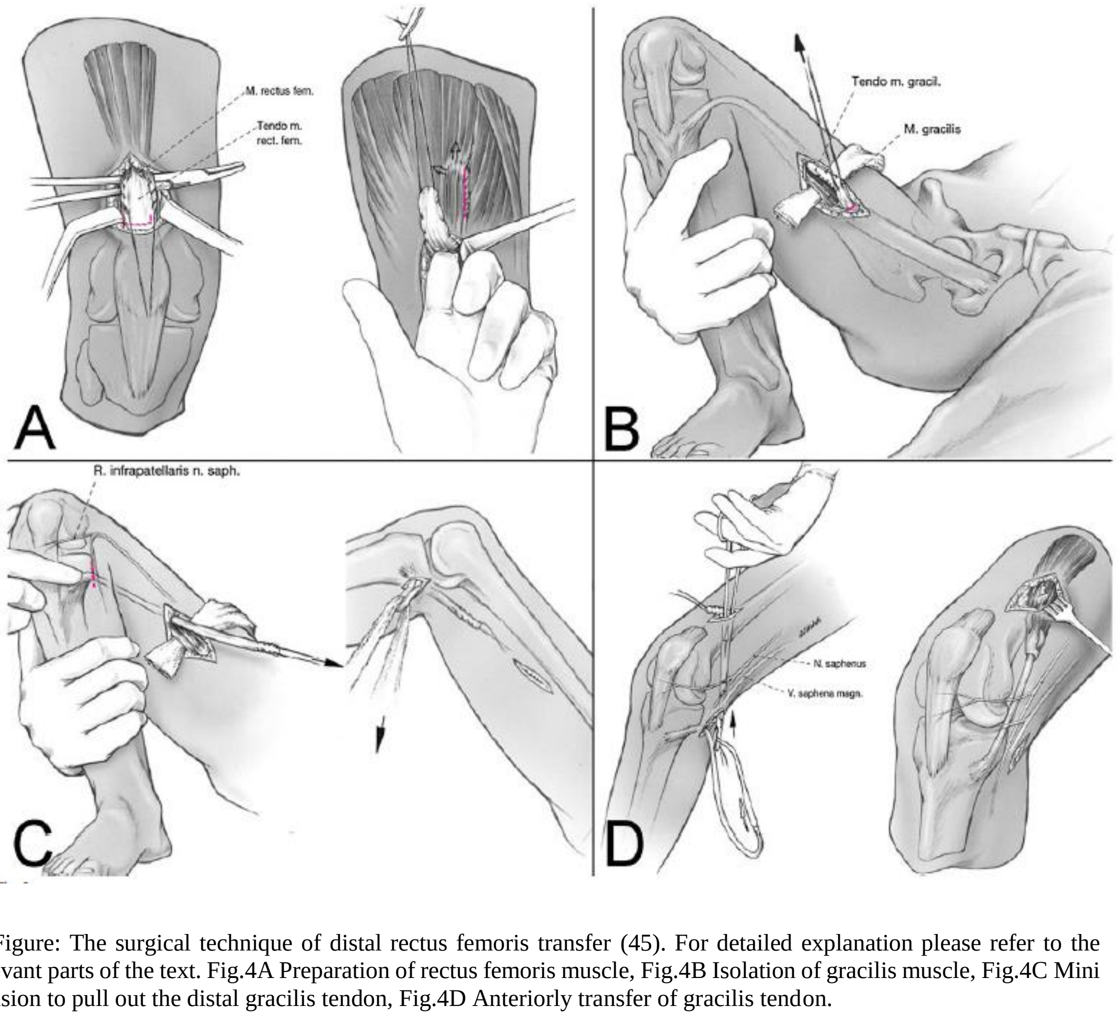
Hamstring lengthening is one type of the MTL, which can be performed by medial and lateral hamstrings to correct flexed knee gait (38, 39). During the operation the patient lies supine and a 3-4 cm long incision is done first medial between the proximal and distal half of the thigh, above the popliteal crease. The semitendinosus and gracilis tendons are then elongated by tenotomy of the proximal intramuscular tendon (Fig.3A). The fractional intramuscular lengthening of the semimembranosus muscle is achieved by making transverse incisions on the aponeurosis of the semimembranosus in single or several levels (Fig.3B). In our studies the gracilis tendon was often used for rectus transfer. If the popliteal angle was greater than 20° after medial hamstring lengthening, biceps femoris muscle was lengthened by an intramuscular aponeurotic lengthening technique.

A  B 

3. Figure: Medial intramuscular lengthening; Fig 3A Semitendinosus muscle, Fig 3B Semimembranosus muscle

1.6.5.1.2. Distal rectus femoris transfer

The treatment of stiff knee gait standardly is the distal rectus femoris transfer (40-44).. (Fig. 4A) The patient lies in the supine position and a 2 cm anterior approach is carried out at the distal part of the thigh, 3 to 4 cm above the proximal patellar pole. After incision of the fascia, the distal rectus tendon is identified and separated from the underlying remaining parts of quadriceps femoris. The tendon is tagged and released as distally as possible (red dotted line in left image). The rectus tendon is then mobilized and further separated from the remaining parts of the quadriceps femoris by digital preparation. Stronger adhesions are released by scissors or scalpel (red dotted line in right image). The fascia is dissected enough to avoid angular deviation of the transfer distally (red dotted line in right image). (Fig. 4B) A medial thigh incision is made to isolate the gracilis muscle. The intramuscular tendon is exposed, tagged, and released proximally (red line) and is successively separated from its muscle belly distally while the knee is flexed. (Fig. 4C) A mini-incision (red dotted line) at the posteromedial knee is used to expose and pull out the palpable distal gracilis tendon. (Fig. 4D) Through the initial anterior approach, a long clamp is passed under the fascia and below the sartorius muscle belly to the mini-incision at the posteromedial aspect of the knee. The gracilis tendon is grasped by the tagging sutures and is transferred anteriorly, where it is sutured to the rectus femoris tendon with moderate tension while the knee is flexed 20 º (45).



4. Figure: The surgical technique of distal rectus femoris transfer (45). For detailed explanation please refer to the relevant parts of the text. Fig.4A Preparation of rectus femoris muscle, Fig.4B Isolation of gracilis muscle, Fig.4C Mini incision to pull out the distal gracilis tendon, Fig.4D Anteriorly transfer of gracilis tendon.

1.6.5.2. Surgical approaches in the central nervous system

Surgical approaches of the central nervous system (CNS) are most commonly done at spinal cord level, with posterior dorsal rhizotomy. It means cutting the dorsal sensory nerve rootlets. There are two types of it: the Peacock approach involves a laminectomy from L1 to L5 with separation of the rootlets as they exit the spinal canal. The consequence of the Peacock technique is a progressive lumbar lordosis (46). The other method is called Fasano technique. It involves a laminectomy from T12-L1 in which the rootlets are separated at the end of the conus. The long-term effect of the Fasano approach is the thoracolumbar kyphosis (47). There is no apparent difference between the outcomes of the two procedures. Although there may be less need of orthopedic surgery after a dorsal rhyzotomy has been performed, others have shown that there definitely is still significant skeletal deformity occurring throughout development possibly necessitating more orthopedic surgery (48). Complications may include hip dysplasia and spine deformities including kypohosis, lordosis, spondylolisthesis and spondylosis (49, 50).

1.6.5.3. Surgical and chemical approaches in the peripheral nervous system

Another way to decrease spasticity is to intervence at the level of the peripheral nerves (48). A lesion can be made either chemically or by physical transection. The chemical agents can be short-acting or long acting anesthetics, as alcohol and phenol. The most commonly sectioned nerve is the obturator to decrease adductor spasticity at the hip. But it should be done only in nonambulatory children, and only the anterior branch of the obturator nerve should be sectioned. Overall the control of spasticity and peripheral neurectomy has a minimal role in the management of spasticity in children with CP.

1.7. Gait

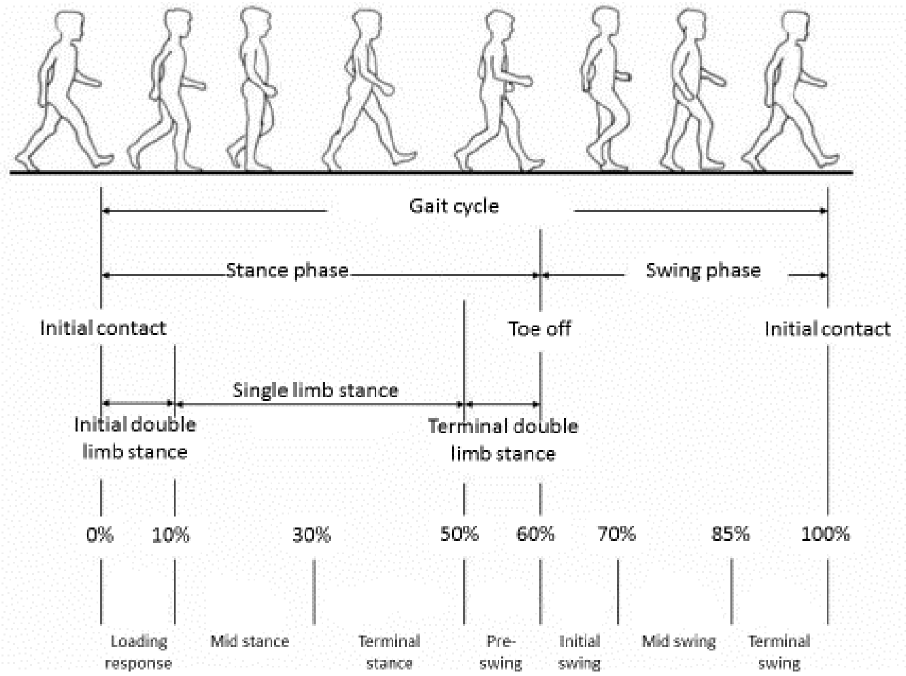
Human gait is a complex interaction between the CNS and the peripheral musculoskeletal system. The gait is a periodically repeating sequence of the lower extremities to move the body forward (51). According to velocity it can be walking, sprinting and running. The stability during walking is kept and the movement of limbs repeated in cycles. The gait cycle is the smallest unit of the gait. This is a single sequence of events between two sequential initial contacts by the same limb.

One gait cycle during walking can be divided into two phases: stance phase (60%) and swing phase (40%) (Fig.5). These phases represent specific functional patterns. There is no specific starting or ending point, with one action flowing smoothly into the next. But because of the best defined event is the floor contact, it has been selected as the start of gait cycle and it is called initial contact. The stance phase begins with initial contact of one limb. A healthy person initiates floor contact with his heel. During stance phase the foot is on the ground. Swing phase follows the stance phase. The foot is in the air for limb advancement. It begins with toe-off that means the foot is lifted from the ground.

Stance phase subdivided into three subgroups according to the sequence of floor contact by the this two feet. At the initial double stance both feet are on the floor after initial contact. The body weight is shared is equally by the two feet. When the opposite limb is lifted for swing begins the single stance. The whole body weight is resting on one limb. It is followed by the third subdivision; the terminal double stance. It begins with floor contact by the other foot and continues until the original stance leg is lifted for swing.

Each gait cycle contains eight functional patterns (52). These are subphases of the gait cycle. The gait cycle (GC) begins with initial contact and takes about 0-2% of GC. It is followed by loading response, namely the initial double stance period that is 0-10% of GC. The single stance period involves two phases: mid stance and terminal stance. Mid stance (10-30%) begins as the other foot is lifted and continues until body weight is aligned over the forefoot, follows the terminal stance. This takes 30-50% GC until the other foot strikes the ground. Stance phase closes with pre-swing (50-60% GC), in other words the terminal double stance. The opposite limb has initial contact on the ground and ipsilateral limb has toe-off. Swing phase subdivides into three phases. Initial swing (60-

73% GC) begins with toe-off and ends when the swinging foot is opposite the stance foot. It follows the mid swing (73-87% GC) till the swinging limb is forward and the tibia is vertical. The final phase of GC is the terminal swing (87-100% GC). It begins with vertical tibia and ends when the foot touches the floor.



5. Figure: Gait cycle (53)

During walking the body functionally divides itself into two units: passenger and locomotor (54). The passenger unit is composed of the head, neck, trunk and arms. It takes 70% of the body weight. The locomotor system is formed by the two lower extremities and the pelvis. Locomotor unit is responsible to move forward the passenger unit (propulsion), stance stability, shock absorption and energy conservation. Stability means a functional balance between the alignment of the body and muscle activity at each joint. There is a point, the center of gravity, that is representative of the weight of that mass. There is a passive stability when the center of gravity of the upper segment is aligned directly over the center of the supporting joint. Its security depends on the quality of the supporting surface and the nature of any external forces. During standing and walking the effect of body weight is identified by the ground reaction force vector. The body weight falls toward to the floor, it creates a force in the floor of equal magnitude but opposite in direction. It can be captured by three dimensional gait analyses. There are three forces that act on the joints: falling body weight, ligamentous tension and muscular activity. The hip and knee can achieve a passive stability when the joints are hyperextended. It is balanced by ligamentous tension and body vector. At the knee there is the posterior oblique ligament. The hip is limited anteriorly by the iliofemoral ligament. In this position the joint are locked by two opposing forces: the body weight vector on one side of the joint and ligamentosus tension on the other. At the ankle there is no similar source of passive stability. The ankle joint is not located at the middle of the foot, but posterior to the center of the foot. The heel lever is much shorter than the forefoot lever, which extends to the metatarsal heads. During walking, the area of support changes from the heel to flat foot and then the forefoot. The body's passive stability changes to dynamic stability. The dynamic stability is modified by continual realignment of the vector to the joints. The center of gravity of the passenger unit is aligned medial to the supporting limb, and the connecting link is highly mobile hip joint. Two preparatory actions need to keep the balance: lateral shift of the body mass and local muscular stabilization of the hip joint.

It is important to establish a clear and common understanding of the terms present. They are founding in the Table 3.

3. Table: Description of biomechanical terms (37)

Term	**Description**
Temporal spatial characteristics	Changes in the body or body segments related to the GC
Gait velocity	Change in distance per unit time of the whole body during gait
Step	The GC of one limb; the distance one foot moves with each GC
Cadence	The number of GC's per unit time
Stride	GC of the whole body that equals two steps
Step width	The distance in the transverse plane of how far the feet are separated during double support.

1.7.1. Three dimensional gait analysis

The origins of the science of gait analysis began in Europe in the 17th century and continued through the early 20th century (55). Today the three dimensional (3D) gait analysis is the gold standard of refined diagnosis and treatment for planning spastic gait disorders. With appropriate systems can be analyzed the kinematic, kinetic, dynamic electromyography and dynamic foot pressure. In modern laboratories the subject reflective markers are fixed on the subject and they walk on a calibrated walkway, while the markers transmit the data from the moving joints to the computers. The resultant joint angles can be viewed within minutes from the end of collection of the data.

However there are still some difficulties to overcome. One of them is the accurate timing of toe-off, while the other is the marker movement over the under lying skeleton on the skin during walking.

Since the inception of the 3D gait analysis it has gained an emerging role in the diagnosis, to plan operations and it is able to give an objective recording after surgery. By now 3D gait analysis´ importance can be evaluated focusing on the relevance of the 3D gait analysis to decision making by the treatment of gait disorders in cerebral palsy patients (56). Some studies showed, that the results are better, if the references of gait analysis are taken into consideration (57, 58). The 3D gait analysis is established as a crucial component of multimodal planning unit (clinic and radiologic examinations, 3D gait analysis).

1.7.1.1.Kinematics

This is a measurement of the displacement of the body segments during gait, usually defined as angular change of the distal segment relative to its proximal articulated segment, or motion relative to a global coordinate system. It is based on dividing the body into 8 segments: trunk, pelvis, two thighs, two shanks and two feet. The principia exist, that reflective or active markers are placed on specific anatomic segments and their motion will be captured by many cameras (Table 4, Fig. 6 and 7). Kinematic motion can be captured by two dimensional (2D) or 3D system. The 2D system is prone to more errors, because it is analyze only the sagittal motion, the frontal and transversal plane will be lost. Because of the loss of information in 2D the 3D gait analysis system is the standard method. Each segment must be defined by a minimum of 3 markers. Each of these markers is imaged by a minimum of two cameras simultaneously. Only this way a marker can be defined in three-dimensional space. The cameras are focused on a fixed place in the room, which is assigned a room coordinate system. These are synchronized to take images at the rate of 60 frames per second. The markers are identified by separate identifiers based on the body segments. These motions can be calculated into clinically defined joint range of motion with a specific software (Plug-in, Vicon®, Oxford Metrics, UK).

The static capture completed with a special device to measure the correct position of the knee (Fig. 7). It is positioned on the medial and lateral femur condyle. Its position gives us the horizontal, sagittal and transversal plane of the knee and the torque of the femur.

4. Table: Position of the reflective markers in Plug-in

Trunk	
C7	*Processus spinosus of C7*
LSHO	*Articulation acromioclaviculare left*
RSHO	*Articulation acromioclaviculare right*
CLAV	*Incisura jugularis*
Pelvis	
SACR	*The half way between two spina iliaca posterior superior*
LASI	*Spina iliaca anterior superior left*
RASI	*Spina iliaca anterior superior right*
Thigh	
LTHI	*Between Trochanter major and Condyle lateralis femoris left*
RTHI	*Between Trochanter major and Condyle lateralis femoris right*
LKNE	*Lateralis condyle of femur left*
RKNE	*Lateralis condyle of femur right*
Shank	
LTIB	*On the lateral side of the tibia between LANK and LKNE*
RTIB	*On the lateral side of the tibia between RANK and RKNE*
LANK	*Malleolus lateralis left*
RANK	*Malleolus lateralis right*
Foot	
LHEE	*Tuber calcanei left*
RHEE	*Tuber calcanei right*
LTOE	*Second metatarsal head left*
RTOE	*Second metatarsal head right*

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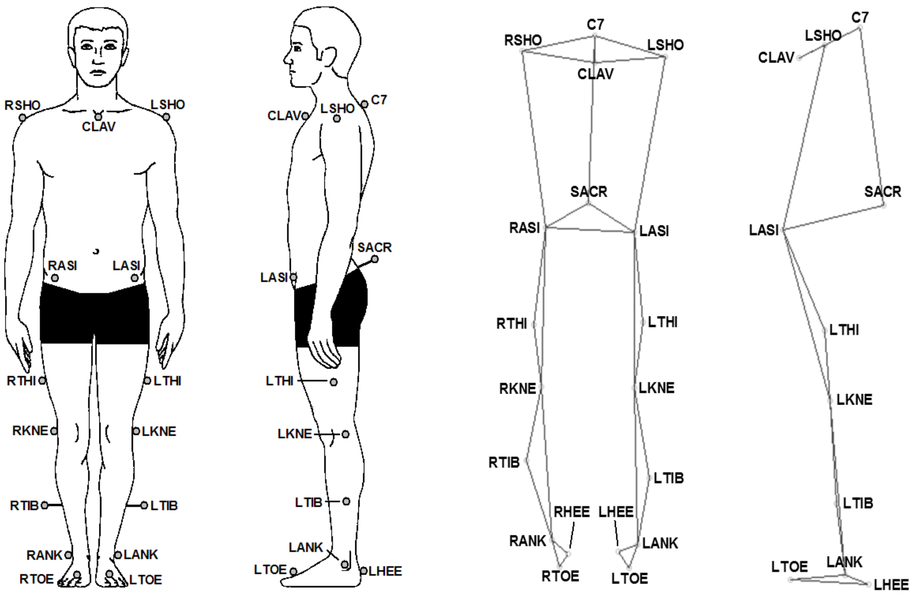
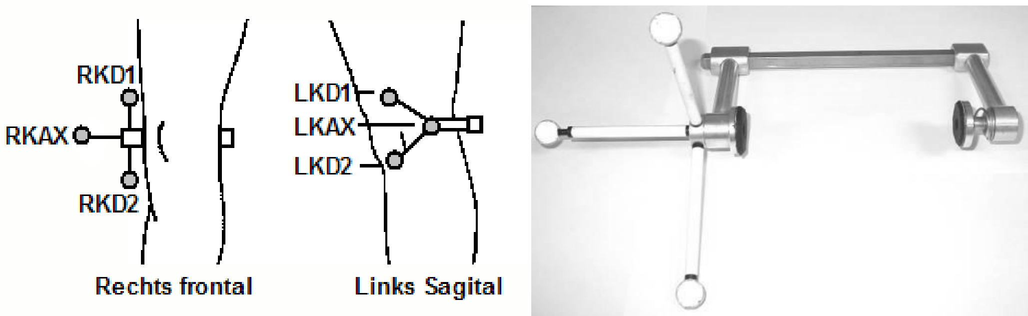


6. Figure: Position of the reflective markers on the body



7. Figure: Positioning of the knee in static capture

1.7.1.2.Time-distance parameters

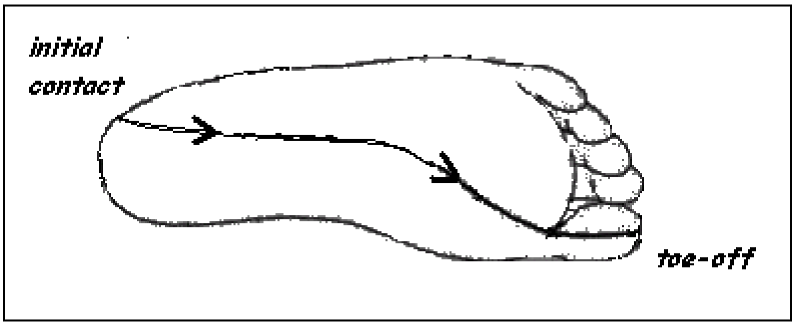
The time-distance parameters were elevated from kinematics data and they give an advice from distance measurement in the entire GC. These parameters are the cadence (number of steps/min), speed (m/s), step length (m) and timing of a step (s). Stride is the interval between two sequential initial floor contacts by the same limb.

1.7.1.3.Kinetics

The measurement of forces at each joint is called kinetic evaluation (37). Net joint forces are indirectly measured as the opposite of the force required to counteract the momentum and ground reaction force (Fig.8). Momentum is measured by assigning each segment a weight and a center of weight, and by the velocity and acceleration of the mass through the use of kinematic measurement. The ground reaction force (GRF) is measured with sensitive and accurate force plates fixed to the floor, over which children walk. At the corner of the force plate there are three sensors measuring the three directions: vertical horizontal and side-ways, as well as moments about each of these axes.

8. Figure: Ground reaction force (37)

Each joint has a direction and distance from the defined center of the joint. By knowing where the joint’s center is in space and the direction of the GRF vector, the moment arm can be calculated. With the knowledge of the moment arm and the GRF vector, the moment generated by the GRF vector can be calculated. The GRF vector cross the force plate, where is the center of the pressure. During stance phase the center of pressure is changing, which can be followed on the foot (Fig.9).



9. Figure: The center change of pressure during stance phase

The moment from the GRF vector is then added to the moment of momentum and the total external joint moment is measured. The muscle, ligaments and bones must create an equal opposite internal force because of the system stability. The joint moment is calculated by the magnitude and direction of the GRF measured from the force plate combined with the momentum component calculated from the kinematic motions of the joint segment.

Moments are typically measured in units of Newtonian meter (Nm), which are then divided by a child's body weight (Nm/kg). Joint powers have units of watts and divided with a child's body weight: W/kg.

1.7.1.4.Electromyography

Electromyography (EMG) is an electrical recording of muscle activity, summation of all muscle fiber action potentials (37). Muscles are stimulated by motor neurons. This stimulation causes electrical activity in the muscle, which can be detected by an electrode. The signal of the electrical activity is diminished due to subcutaneous fat and skin. The EMG has to be correlated to the gait cycle by synchronizing the EMG to the kinematic measurements (Tabl.5).

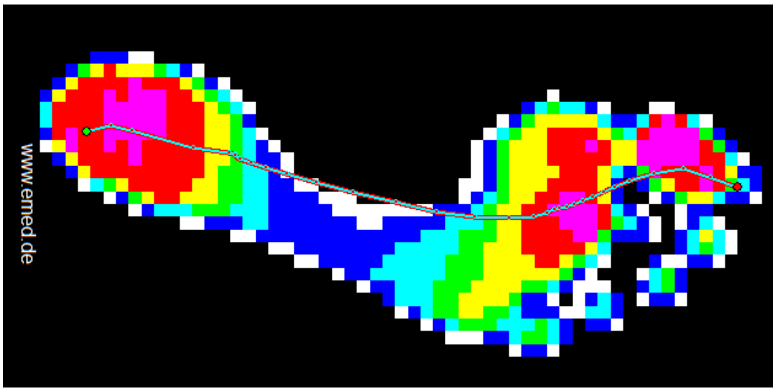
5. Table: Clinical definitions of EMG activity

Terminology	Definition
Early onset	*Activity of the muscle begins before the normal onset*
Prolonged	*Muscle activity continues past the normal cessation*
Continuous	*The muscle is always on with no turn-off time (constant activity may be hard to distinguish from no activity that generates background noise).*
Early off	*Early termination of the muscle activity*
Delayed	*Onset of muscle activity is later than normal*
Absent	*No muscle activity, which can be hard to separate from continuous activity.*
Out of phase	*The muscle is active primarily during the time it would normally be silent and is silent when it should be active.*

Utilizing EMG besides the 3D gait analysis we are able to address the normal muscle pattern or its deviations from the physiological standard. Frequently analyzed muscles are: *rectus femoris, biceps femoris, vastus lateralis, semimembranosus, gastrocnemius lateralis, soleus, tibialis anterior.*

1.7.1.5.Pedobarograph

The measurement of the pressure distribution on the sole of the foot is called a pedobarograph (Fig.10). This allow us to diagnose planovalgus or equinovalgus foot deformity in CP children. The children have to walk over the measurement plate without targeting the plate. The information will be reliable and it is the best way currently to monitor childhood foot deformities. The test is quick and easy to obtain information.



10. Figure: Pedobarograph

1.7.1.6.GGI

The Gillette Gait Index (GGI) was defined by Schutte et al. in 2000 (59). This index is used to measure pathologic gait severity and assess therapeutic outcomes in children with CP. It incorporates 16 clinically important spatial, kinematic and temporal parameters (Tabl.6), and estimates the deviation of a patient's gait from a normal gait pattern. It was found, that the mean index for the control group is the lowest (15.7) and the quadriplegic patients have the highest (491.0). There was no overlap between index values for the control subjects and the individuals with gait abnormalities. With this index the surgical outcomes and postoperative changes can be easy quantified.

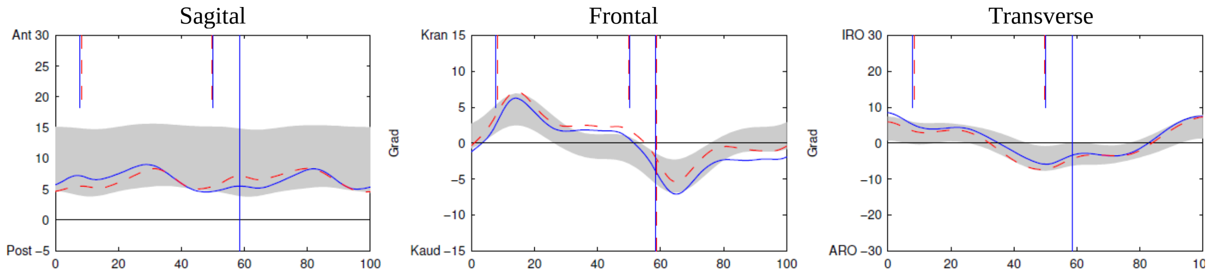
6. Table: 16 parameters of GGI

Time of toe off	Peak abduction in swing
Walking speed	Mean hip rotation in stance
Cadence	Knee flexion at initial contact
Mean pelvic tilt	Time of peak knee flexion
Range of pelvic tilt	Range of knee flexion
Mean pelvic rotation	Peak dorsiflexion in stance
Minimum hip flexion	Peak dorsiflexion in swing
Range of hip flexion	Mean foot progression angle

1.7.2. Normal gait

1.7.2.1.Pelvis

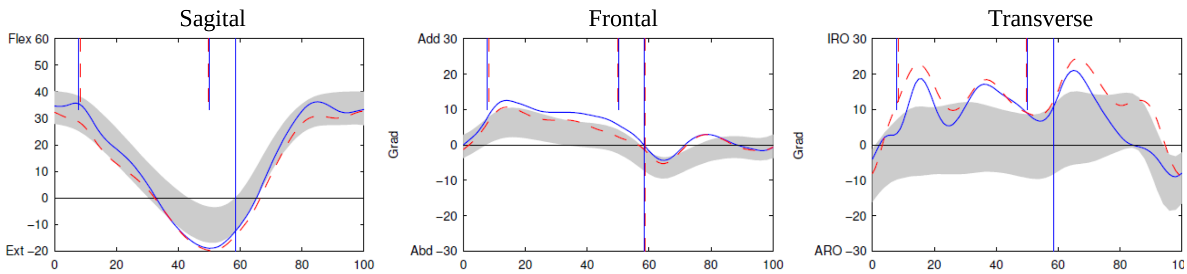
Pelvis belongs to passenger unit, but its function is special: it connects the two hips with each other (60). Throughout the GC the pelvis deviates in all three planes: sagittal, coronal and transverse. The site of action is the supporting hip joint. All motions are very small (Fig.11): anterior/posterior tilt 4°, contralateral drop/rise 4°and posterior/anterior rotation 10°. But it increases with increasing speed of walking. Two muscle groups were identified, which are responsible to control the pelvic movement: abductors and extensors of the hip. From the abductors two muscles are specifically involved: gluteus maximus and medius.



11. Figure: Pelvismotion. Grey area represents the median and the standard error of the mean of control group. The blue continuous and red dashed line is the left and the right leg of a representative subject respectively.

1.7.2.2.Hip

Hip is the junction between the passenger and locomotor units (61). During a normal stride the hip moves through only two arcs of motion: extension and flexion (Fig.12). Its average takes out 40° (62). The peaks of hip movements on sagittal plane are 10° for extension and 30°for flexion. There is a minimal movement on the coronal plane as the unloaded side of the pelvis follows the swinging limb. At initial contact, the hip adducted about 10°and it decrease to 5°during loading response. At initial swing a relative hip abduction occurs with 5° (63). The total arc of transverse hip motion averages 8°. If it is added to pelvic rotation, the total thigh rotation is 15°. In stance the primary muscles which control the hip, are the extensors and abductors (biceps femoris, semimembranosus, semitendinosus, adductor magnus, gluteus maximus, gluteus medius, gluteus maximus and tensor fascia lata), while during swing it is the flexors (adductor longus, rectus femoris, gracilis, Sartorius and iliacus) .

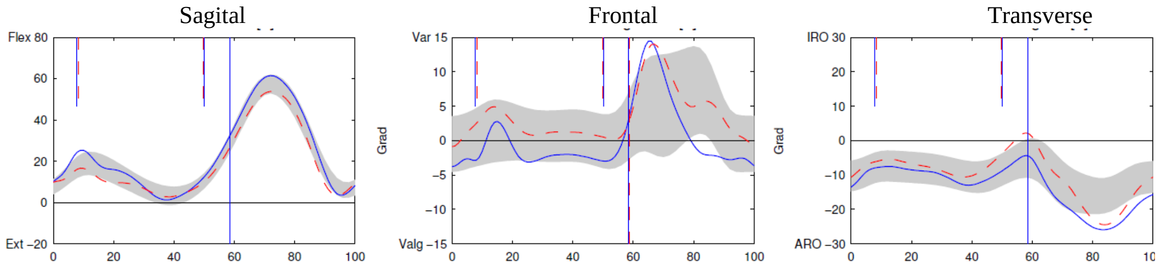


12. Figure: Hipmotion. Grey area represents the median and the standard error of the mean of control group. The blue continuous and red dashed line is the left and the right leg of a representative subject respectively.

1.7.2.3.Knee

The flexibility and stability of the knee are major factors in the normal pattern of walking (64). In stance the knee gives the basic stability of the limb and during swing it is responsible for the leg's freedom to advance. There are some two joint muscles that also control the moving of ankle and hip.

The normal knee motion has a big range in sagittal plane (0-70°) during normal walking (Fig.13). The differences between studies of knee motion are related to variations of walking speed, subject individuality and the landmarks. At initial contact the knee is flexed about 5°. At the first half of the stance phase the knee is flexed to 20-25°. This is the time when the flexed knee is under maximum weight-bearing load. At the second part of the mid stance, the knee extends. Shortly before the knee slowly begins to flex again in terminal stance, the knee procure the maximal extension of it with about 3°. During terminal stance the knee begins its second flexion. At mid-swing the knee is flexed to the maximum (60-70°). In terminal swing the knee begins to extend rapidly. It continues till full extension (5°) is gained (65). In the transverse plane the knee shows about 9° rotation during the GC. It begins with external rotation at the end of stance, and from the toe-off it continuous with internal rotation during swing and loading response. In the coronal plane there are abduction and adduction. During stance phase the motion is abduction and during swing the knee returns to adduction with 8°(66). The thirteen muscles contributing to knee control contract at selected intervals within the GC. Only five muscle are unique to the knee. The extensors are the three vasti heads, and two flexors are the popliteus and short head of the biceps femoris. All the other muscles control either hip or ankle motion. One of the two joint muscles is rectus femoris which contributes to knee extension. The gluteus maximus provides a knee extensor force through its iliotibial band insertion. The three hamstring muscles (semimembranosus, semitendinosus and biceps femoris long head) are two-joint muscles (10). These are known as knee flexors. The gastrocnemius participates in knee flexion, while principally acting on the ankle. The gracilis and sartorius also contribute to swing phase knee flexion.



13. Figure: Kneemotion. Grey area represents the median and the standard error of the mean of control group. The blue continuous and red dashed line is the left and the right leg of a representative subject respectively.

1.7.2.4.Ankle

Ankle is the junction between tibia and foot. There are two single axis joints to provide three-dimensional mobility: subtalar and tibiotalar articulations. During a GC four arcs of ankle motion occur: two plantar flexions (PF) and then two dorsiflexions (DF) (Fig.14). In stance PF, DF and PF follow each other. During swing the ankle only dorsiflexes (65, 66). The whole range of ankle motion extends about 30°. Initial contact by the heel occurs with the ankle at neutral or minimal plantar flexed (0-5°). The maximal dorsiflexion takes place in terminal swing with about 10°. The maximum of plantar flexion is at initial swing (20°).

The ankle controlling muscles function either dorsiflexiors (tibialis anterior, extensor halluces longus and extensor digitorum longus) or plantar flexors (soleus, gastrocnemius, tibialis posterioir, flexor digitorum longus, flexor halluces longus, peroneus longus and peroneus brevis.

14. Figure: Anklemotion. Grey area represents the median and the standard error of the mean of control group. The blue continuous and red dashed line is the left and the right leg of a representative subject respectively.

1.7.3. Spastic diplegic gait

The cerebral palsy patients have been found to exhibit several common gait abnormalities. With respect to the knee, there are four common pathologic patterns seen in ambulatory patients with spastic diplegic-type cerebral palsy. Despite three dimensions of knee motion, the common gait abnormalities in cerebral palsy occur in the sagittal plane. Coronal plane abnormalities appear less frequently and are usually attributable to abnormalities at other joints, such as the extreme femoral anteversion or valgus instability of subtalar joint. Following knee abnormalities are just secondary problems. Transverse plane abnormalities at the knee are also secondary phenomena, most often attributable to femoral or tibial torsional deformities (67). The gait abnormalities in cerebral palsy patients lead to contractures across joints and muscle spasticity. In 1993, Sutherland and Davids classified the common gait abnormalities of the knee in cerebral palsy into four types: jump, crouch, recurvatum, and stiff (67).

1.7.3.1.Crouch knee

Increased knee flexion during the stance phase of gait called flexed knee (crouch knee) gait, with variable alignment in swing phase (Fig.15) (68). Hamstring spasticity with resultant contracture has been identified as one of the main factors leading to a flexed

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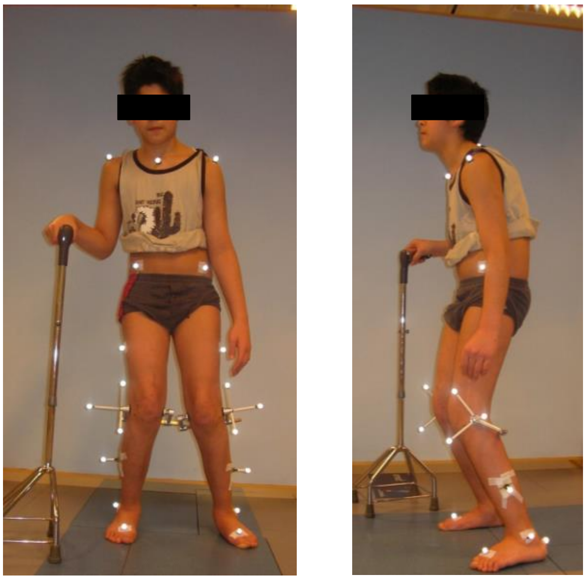
knee gait (69-74). Later investigations have shown that increased external tibial torsion, instability of the foot, and quadriceps weakness can cause or aggravate flexed knee gait (75-78). Knee kinematics on sagittal plane shows an increased knee flexion of at least 30°during stance phase. Decreased knee extension in terminal swing will be mostly presented. The peak knee flexion in swing (pKfSw) is over 50°, causing shortened stride length, resulting in a decrease in velocity. On the EMG in stance phase prolongation of quadriceps and hamstring activity are usually presented (67). During stance phase the increased knee flexion results a continuous flexion moment about the knee, leading to excessive demands on the quadriceps (79).



15. Figure: Crouch knee

1.7.3.2.Stiff knee

Stiff-knee gait is a common gait abnormality in patients with spastic diplegic cerebral palsy (68). Decreased knee flexion during swing phase leads to foot clearance problems, reduces gait velocity, and reduces step length. According to Rodda et al. (80) stiff-knee gait shows a decreased knee excursion throughout the whole gait cycle of <30°, whereas

Sutherland et al. described it as delayed and decreased pKFSw phase with diminished total knee motion (81).

The spasticity of the rectus femoris muscle is the primary cause of stiff-knee gait (42, 72, 82, 83). Rectus femoris is pathologically active during swing phase, counteracting the knee flexion necessary for foot clearance and step length (42, 67, 72, 82, 83).

1.7.3.3.Jump knee

The name is telling, because the subject is jumping up and down. The patients are usually younger than those with crouch knee. There is increased knee and hip flexion in early stance phase (at least 30°), and extend variable in mid and terminal stance (10-20°), but never reach normal knee extension. In late swing there is a decreased knee extension (67). The ankle is in equinus, especially in late stance by increased tone in triceps surae. The movement of pelvis during GC is either normal or tilted anteriorly (80). Hamstrings show an increased tone, the hip adductors and in flexors mild contracture is present. Quadriceps weakness can be occasionally seen. During stance phase, hamstrings present overactivity.

In case of younger children whit minimal or no contracture present, selective dorsal rhizotomy may be considered (84). Multilevel botulinum toxin A injection to the psoas, hamstrings and calf can be suggested in children with jump knee (85). In the third level, when there are contractures, simultaneous bilateral single event multilevel surgery on the muscles can be considered: psoas, hamstrings and calf (85). The GRF is anterior to the knee, leading to overactivity in plantar flexors and knee extensors. A posterior leaf spring or hinged ankle foot orthosis restricts excessive plantar flexion and aids movement into dorsiflexion, thereby realigning the GRF and normalizing the plantar flexion knee-extension couple (86).

1.7.3.4.Recurvatum knee

The etiology of recurvatum knee can be triceps surae contracture or overactivity in mid or terminal stance, leading equinus in ankle and excessive lengthening or transfer of hamstrings (9). It is characterized by increased knee extension in mid or terminal stance with variable knee motion in swing phase. In physical examination a hyperextension in knee ROM can be present. This hyperextension can lead to varus-valgus deformity, especially in older children. The equinus contracture often presents significantly increased tone and clonus. The knee motion shows a hyperextension in mid and terminal stance phase, the pKFSw is delayed and diminished. The EMG demonstrates an overactivity of the triceps surae and elongated quadriceps activity in stance phase. The hyperextension leads decreased velocity by limiting stride length. In the kinetics, it exhibits an excessive flexor moments throughout stance phase.

2. Purpose

2.1. Development of knee function after hamstring lengthening as a part of multilevel surgery in children with spastic diplegia

Lengthening of the hamstrings is widely considered to be the standard surgical procedure for the correction of increased knee flexion (26, 38, 87, 88). The hamstrings can be lengthened with use of an open or a percutaneous technique (88-91). Satisfactory short-term results with improved knee extension during stance phase have been shown in different studies (41, 87, 88, 91). However, outcomes have been inconsistent; many patients have had improvement, whereas other patients have had only little benefit or even worsening (41, 88-92). After short term follow-up, some authors have reported increased pelvic tilt and a high prevalence of genu recurvatum as a consequence of overcorrection (38, 88, 91). It is an interesting question, whether is there any different in the appearance of the anterior pelvic tilt between medial and combined medial and lateral hamstring lengthening in short and long-term, however it was already analyzed short-term by Kay et al. (88). Evidence for the effectiveness of hamstring lengthening to correct flexed knee gait in spastic diplegia is scant because of small and inhomogeneous case series, different surgical techniques, and short follow-up. As a result, hamstring lengthening currently is viewed as controversial. A major problem is that, to our knowledge, there have been no long-term studies investigating the effects of hamstring lengthening in skeletally mature patients who were managed in childhood. Therefore, the long-term results in adolescents and adults who had had hamstring lengthening as a part of multilevel surgery in childhood were investigated in one of our study. Our hypothesis was that correction of flexed knee gait seen at short-term follow-up is not maintained in the long term.

2.2. Long-term results after distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff knee gait in spastic diplegic cerebral palsy

The distal rectus femoris transfer (DRFT) is the standard surgical procedure for the treatment of stiff-knee gait. The tendon transfer can be occurred to medial hamstrings or to lateral hamstring or iliotibial tract (72, 83). It aims to improve knee flexion in swing and pKFSw for foot clearance. The criteria of DRFT in the literature are inconsistent (67, 72, 93-98). Some studies have noted good initial results, with an improvement in pKFSw and knee flexion in swing following DRFT (93-95). Nevertheless, some authors have suggested that DRFT does not generate a knee flexion moment, although the capacity of knee extension is diminished (96-98). Other authors have described no significant increase of pKFSw but an increase of total knee motion (39, 43, 99). Summarized, the results after DRFT are inconsistent. In addition, it is debatable whether patients with severe flexed knee gait who show highly increased knee flexion throughout stance phase and a normal or even increased pKFSw phase should receive a prophylactic DRFT to preserve pKFSw after correction of flexed knee contractures by concomitant procedures such as hamstring lengthening or femoral extension osteotomy. There are only a few studies, which have longer-term outcomes after DRFT. Saw et al. showed a significant improvement in pKFSw phase 1 year after surgery, but this improvement in knee flexion in swing decreased and was associated with a loss of knee extension during stance phase 4.6 years postoperatively (93). Moreau et al. also reported significant improvements three years after surgery (94). However, these studies were based on relatively small numbers, and both included patients with different types of cerebral palsy. Furthermore, recruitment was done from different treatment centers that used different surgical techniques of tendon transfer, and the time to the last follow-up was inconsistent, ranging from 0.9 to 6.7 years after surgery. Long-term studies with adequate patient numbers, homogeneous patient groups, and a follow-up interval of more than five years, when growth is expected to be finalized, are missing. The purpose of our study was the evaluation of long-term results (mean, 9 years) in adolescents and adults with diplegic cerebral palsy who were treated with DRFT as a part of single-event multilevel surgery in their childhood. Since the indication for DRFT differed between patients with decreased pKFSw and those with normal or increased pKFSw, in which the DRFT was done as a prophylactic procedure to preserve pKFSw after the correction of severe flexed knee contracture in stance phase, these two groups were analyzed separately.

2.3. Does proximal rectus femoris release influence kinematics in patients with cerebral palsy and stiff knee gait?

The proximal part of the rectus femoris is believed to play an important role in pelvic tilt and hip flexion (81, 100). Hence, to treat hip flexor contracture and increased anterior pelvic tilt (APT), proximal rectus femoris release can be considered (100) and still is performed sometimes, although few studies have investigated its effects (81, 100). However, McMulkin et al. found no relevant influence of proximal rectus release on the knee in patients in whom DRFT was not performed (100). Sutherland et al. compared DRFT and proximal rectus release concerning the effect on knee kinematics and found significantly less effect for the group in which a proximal rectus release was performed (81). The main effects of DRFT on stiff knee gait are seen mainly as consequences of its distal release and not as an augmentation of the knee flexor muscles (101). Fox et al. reported a potential indirect effect of the hip flexion on the knee induced by the rectus femoris (101). In the literature, there are no reports on the outcome if both ends of the rectus femoris are surgically treated simultaneously. The question therefore arises regarding whether an additional proximal rectus release affects knee function when done simultaneously with DRFT. The purpose of our study was to investigate the effects of an additional proximal rectus release on the knee kinematics when done in combination with DRFT. We sought to determine whether an additional proximal rectus release affects knee and pelvic kinematics when done in combination with DRFT; specifically, we sought to compare outcomes using the (1) range of knee flexion in swing phase (2) knee flexion velocity and (3) peak knee flexion in swing, and (4) spatiotemporal parameters between patients treated with DRFT, with or without proximal rectus release. The effects on (5) APT in both groups were compared.

2.4. Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendinous lengthening in children with spastic diplegia

For the correction of flexed knee gait, various surgical strategies have been employed. One of the standard treatment is the hamstring lengthening (9, 26, 38, 57, 87, 88, 91). There are various studies about hamstring lengthening with different short-, mid- and long-term results with incoherent results (9, 26, 38, 57, 87, 88, 91). It can be explained by the influence of the lengthening of biarticular muscles on both adjacent joints (102). To control the biarticular muscles in patients with cerebral palsy is more difficult than the monoarticular muscles (34, 103). The surgical elongation of these muscle tendon unit may add to muscle weakness after surgery (104). The muscle weakness in specific muscle groups lies primary on brain damage in cerebral palsy patients and it will be aggravated by surgical elongation.

There are some studies, which take a conversion of biarticular muscles to monoarticular muscles (34, 105-107). But there is a lack of long-term reports comparing the conversion of biarticular to monoarticular muscles (CBM) results with those of conventional MTL by means of 3D gait analysis. Therefore the purpose of this study was to compare these two groups and investigate its long-term effects.

The presence of anterior pelvic tilt by hamstring lengthening is commonly mentioned in the literature (38, 88, 91). One of our investigation was to compare the outcomes of hamstring lengthening and hamstring transfer to the distal femur. Are there better outcomes of anterior pelvic tilt or long-term recurrence of flexed knee gait?

3. Methods

3.1. Overview

The patients were recruited from CP ambulant of Department for Orthopaedic and Trauma Surgery, Heidelberg University Clinics. The ambulatory patients were selected for surgical correction and they regularly receive a standardized evaluation, which includes conventional 3D gait analysis and clinical examination, both before surgery and at regular follow-up examinations.

The subjects gave informed consent to participate and the studies were approved by the institutional ethics committee.

3.1.1. Clinical examination

The gait of patients were observed and characterized to document the deformities, the ability and the gait asymmetry. The patients were clinically examined standing, siting, as well as in ventral and dorsal position. In this connection were determined the axes deviation of horizontal, sagittal and transversal plane, instability of hip, knee, ankle, subtalar and Chopart-joints as well as difference in leg length. The ROM of the hip, knee and ankle joints were examined according to neutral-0°method, to document the muscle shortening (108). For detecting hip flexion contracture, named after Hugh Owen Thomas, the Thomas test is used. It is performed in dorsal position, one leg is adequately flexed to eliminate lumbar lordosis, and the angle between the longitudinal axis of the thigh and a horizontal line is defined as a hip flexion contracture (109). Furthermore the arbitrary muscular strength according to MRC-scale and the grad of spasticity (modified Ashworth-scale, modified Tardieu-scale, Duncan-Ely test and popliteal angle) were analyzed (7, 110-114). The Duncan-Ely test was demonstrated in ventral position of the patient. The knee was rapidly flexed and when ipsilateral hip rise occurred, the test was positive, otherwise negative (112, 113). The popliteal angle is determined by measuring the angle that the tibia subtends with the extended line of the femur when the ipsilateral hip is flexed to 90° and the knee of the limb under examination is maximally passively extended (114). If the pelvic position will be neutralized by a Thomas test on the opposite side, could be seen the real shortening of the hamstrings.

3.1.2. 3D gait analysis

A conventional, marker-based Vicon® 3D-motion captured system (Oxford Metrics, Oxford, UK) was used to track the 3D positions of 25 reflective markers during walking according to Plug-in Gait marker set (Oxford Metrics) (115). The kinetic, kinematic and EMG data were collected simultaneously during level walking over a 7 m walkway (Fig.16). Before 2002, examinations were carried out with use of a 50 Hz six-camera Vicon® 370 system. Later a 120 Hz twelve-camera Vicon® 612 system was utilized and equivalency of both systems was carefully checked. The cameras were focused on the walkway and they captured the motion of reflective markers. Before all of the measurements the cameras are newly calibrated. Two force plates (Kistler®, Winterthur, Switzerland) were used to collect the kinetic data. For the collection of EMG data an 8-channel device (Noraxon®, Vienna, Austria) with bipolar surfaces adhesive electrodes (Blue Sensor, Ambu Inc., Glen Burnie, MD, USA) according for the SENIAM guidelines was used (116). Pre-amplification of the resulting EMG signal was done by using of Biovision EMG apparatus (Biovision Inc., Wehrheim, Germany). EMG data were digitized using a 16-bit A/D card with the sample frequency of 1080 Hz (117). The EMG signals were fully commutated off-line and the linear envelops were calculated at the cut-off frequency of 9 Hz (118). The EMG amplitudes were normalized to the mean value for each muscle of each step and subject respectively. Time normalization of EMG data was performed for stance and swing phase independently to account for different gait cycle timing between subjects with CP and the reference group of 20 typically developing age-matched children.

16. Figure: Gait analyzes laboratory

The kinematic, kinetic and EMG data's are transmitted by a charging amplifier to an A/D-converter. It converts the data to digital and sends to the computer. All of the data were integrated into a custom-made database. For each patient, 10 to 15 trials were recorded and the data of at least five representative strides of different trials were averaged. All examinations were performed by a specially trained physiotherapist and a study nurse with special education in neurodevelopmental disorders and long-term experience in working with children with CP.

All patients underwent standardized single-event multilevel surgery, including osseous and soft tissue procedures. The surgical procedures were performed according to specific criteria based on clinical examination and gait analysis.

Subjects and/or families or caretakers gave informed consent to participate in a study, which was approved by an institutional ethics committee.

3.1.3. Statistics

Data are presented as mean ± SD, n represents the number of subjects. Comparisons between two groups were performed with unpaired two-tailed Student's t test and multiple group comparisons at different time points were performed applying one-way ANOVA followed by Bonferroni- post hoc test. $p<0.05$ was considered to be statistically significant. Where unique statistical analysis was used is mentioned in the text at the corresponding figure, but it will be summered, what kind of tests and statistical programs were applied in the whole study.

Two types of statistical programs were used: PASW Statistics 18 (SPSS, Chicago, Illinois) and the Prism 5 (GraphPad Software, La Jolla, CA, USA). Various tests were applied in the papers: McNemar, Levene and t tests by Bonferroni and Tukey post hoc tests.

3.2. Development of knee function after hamstring lengthening as a part of multilevel surgery in children with spastic diplegia

For this study, all diplegic patients with flexed knee gait, treated with medial (MHL group) or combined medial and lateral (CHL group) hamstring lengthening in the context of multilevel surgery in childhood between 1996 and 2003 were selected from the gait lab database. For this criterion 155 patients were identified. But for inclusion for this study, the patients had to have at least two standardized postoperative clinical examinations and 3D gait analysis at 1 year and 2 to 4 years after surgery. For these criteria 94 children were found in our database. These 94 children were scanned for exclusion criteria and afterward reinvited for a third long-term follow-up examination at least 6 years after surgery. The exclusion criteria were previous orthopaedic surgery (8 patients), dyskinetic cerebral palsy (5 patients), botulinum-toxin-A injections in the six months prior SEMLS (5 patients), severe mental retardation (3 patients) and loss of walking mobility (2 patients). Three patients were also excluded, because they have had revision surgery for hamstring lengthening or other secondary procedures between the examinations, which potentially may have had influenced the results. After the exclusion of patients, 68 patients remained eligible for reevaluation. 39 children (age at surgery: 10.3 ± 3.5years) could be included in this study.

Static findings including range of motion and popliteal angle and dynamic findings obtained by conventional instrumented 3D-gait analysis were evaluated at four points in time (114, 115): before (E0), one year (E1), 2-4 years (E2), and 6-12 years (E3) after surgery. Demographics and results of GMFCS testing and baseline sagittal pattern are displayed in Table 7 (12, 13).

7. Table: Demographic data and functional baseline parameters (9)

Parameters	E0	E1	E2	E3
Sex: ♀ = 13 ♂ = 26				
Time of follow-up (years)		1.0 (± 0.2)	3.1 (± 1.0)	8.1 (± 1.8)
Age of patients (years)	10.2 (± 3.5)	11.3 (± 3.5)	13.5 (± 3.3)	18.5 (± 4.5)
GMFCS I	6	11	14	13
GMFCS II	21	11	16	13
GMFCS III	12	17	9	13
Preoperative sagittal gait pattern (No. of limbs)				
Group I (true equinus)	0			
Group II (jump knee)	33			
Group III (apparent equinus)	12			
Group IV (crouch)	33			

Legend: GMFCS: gross motor function classification scale. **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively

Hamstring lengthening was considered in diplegic patients with increased knee flexion at initial contact (at least 20°) and/or in mid-stance (at least 10°) observed in 3D gait analysis in combination with at least 30° of knee extension deficiency in popliteal angle test and/or any knee contracture.

All of the patients received standardized SEMLS, including bony and soft-tissue procedures (Tabl.8). Medial hamstring lengthening was performed at the beginning of the surgery. The intra-operative goal for correction was an intra-operative popliteal angle of 20°. The surgical method of hamstring lengthening is written in the chapter 1.6.5.1.1.

8. Table: Number of surgical procedures performed in single event multilevel surgery (9)

Procedures	All (n=78 legs)	MHL-Group (n=61 legs)	CHL-Group (n=17 legs)
Medial hamstring lengthening	78	60	18
Lateral Hamstrings lengthening	18	0	18
Psoas over the brim	14	7	7
Proximal rectus femoris recession	36	30	6
Adductor longus recession	10	7	3
DRFT	71	56	15
Calf muscle lengthening	54	45	9
Soft-tissue Foot (split or complete posterior tibial tendon transfer, split anterior tibial tendon transfer)	18	14	4
FDO	48	38	10
TDO	5	2	3
Bony foot stabilization (Evans, Grice, Chopart or triple fusion, CC-Distraction)	41	31	10

Legend: MHL (medial hamstring lengthening); **CHL** (combined media and lateral hamstring lengthening); **DRFT**: distal rectus femoris transfer; **FDO**: femoral derotation osteotomy; **TDO**: tibial derotation osteotomy; **Evans**: calcaneal lengthening osteotomy; **Grice**: extra-articular subtalar fusion; **CC-Distraction**: calcaneo-cuboid-joint distraction arthrodesis

Postoperative management consisted of early mobilization with immediate weight-bearing. During mobilization epidural analgesia was used to decrease postoperative pain. In those patients, who have had bony procedures, weight-bearing was done 3 to 4 weeks after operation. In patients with residual knee contracture, controlled serial thigh casting was used after surgery to achieve full knee extension within 5 to 7 days. All patients were fitted with night-bearing thigh splints were used for at least 6 months to maintain knee extension.

The types of flexed knee gait were classified for all patients according to Rodda et al. (80) (Tabl.7).

Statistical analysis of the selected parameters from the clinical examinations and 3D gait analysis was done only for the left legs of the patients, as indicated by significant right-left correlations ($p<0.05$). Furthermore, subgroup analysis was performed for the MHL and CHL groups. Descriptive statistics were used for basic statistical analysis. One-way repeated measures analysis of variance was used to show time-changing effects. To assess significant changes in the prevalence of adverse effects (genu recurvatum, increased pelvic tilt) at the 3 examinations after surgery, the McNemar test was applied. Statistical analysis was done using PASW Statistics 18 (SPSS, Chicago, Illinois).

3.3. Long-term results after distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff knee gait in spastic diplegic cerebral palsy

85 ambulatory children with spastic diplegic cerebral palsy (GMFCS I, II and III) were selected for this study, who underwent distal rectus femoris transfer (DRFT) as a part of SELMS before 2004. The inclusion criteria were for this study: an age of 6-16 years at the time of surgery, a standardized follow-up examination at one year after surgery, and a positive pre-operative Duncan-Ely test (112, 113). After next criteria some patient were excluded from this study: previous orthopedic surgery in lower extremity (7 patients) or botulinum toxin A injection less than six month prior to SEMLS (8 patients) and patients with dyskinetic type of CP (3 patients). After the exclusion of patients, 67 patients remained eligible for reevaluation. They were invited for a long-term follow-up examination at least 6 years after orthopedic surgery. 9 of these children could not be evaluated because of moving (7 patients) or had declined to return follow-up (1 patient) or to participate in the study (1 patient) because of personal reasons, and 5 patients were excluded because of relevant secondary surgery (1 hip reconstruction surgery, 2 revision femoral derotation osteotomies, 2 revision hamstring lengthening procedures). Altogether, 53 patients could be participated in this study (Fig.17).

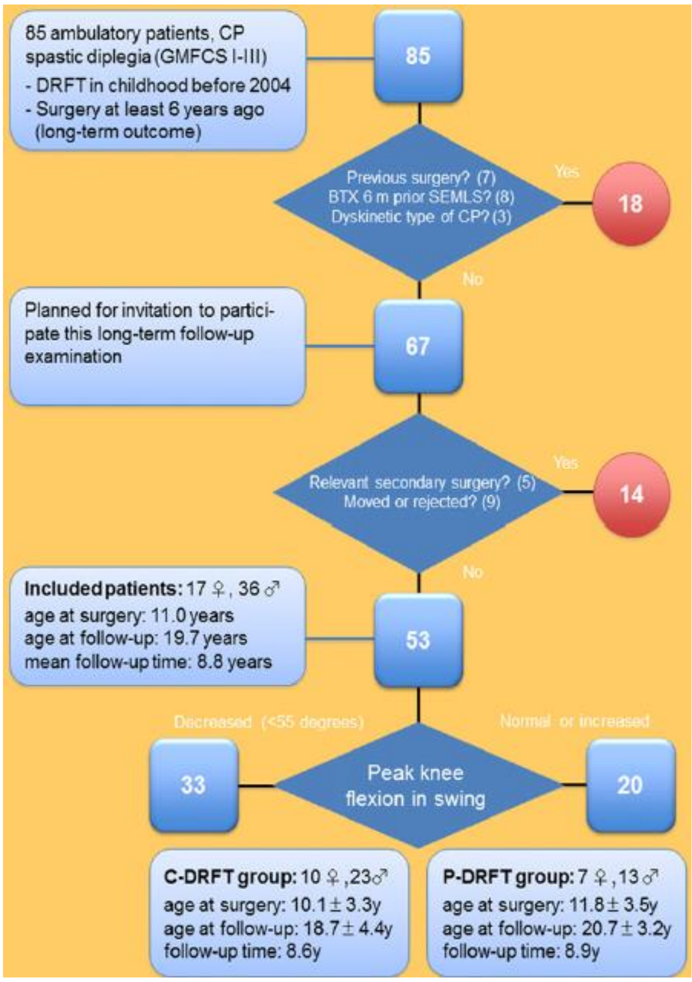
They were reevaluated (17 female and 36 male with a mean age at the time of surgery of 11.0 ± 3.4 years). After our standardized protocol, the patients were examined before surgical procedure and at 1 year and 6 to 14 years (8.8±2.3 years) postoperatively.

The standardized DRFT was done to the gracilis tendon (98 legs in 49 patients). When the gracilis was too thin, DRFT was done to distal tendon of the semitendinosus muscle (8 legs in 4 patients). The surgical technique is described in the chapter 1.6.5.1.2.

After operation the patients were early mobilized with immediate weight-bearing transfers and subsequent walking within the first week after SEMLS. Knee-ankle-foot orthosis in knee extension were applied at night for six months. If the patients have had any bony procedures, this schedule was modified accordingly.

DRFT is generally indicated, if the patients have a positive Duncan-Ely test, a decreased knee flexion in swing and decreased peak knee flexion in swing (pKFSw) as well as pathologically increased activity of the rectus femoris muscle with dynamic EMG (39, 42, 43, 67, 72, 83, 93-95, 99). The patients were divided into two groups: C-DRFT and P-DRFT. In the C-DRFT group the DRFT was done as a correction of knee flexion in swing and peak knee flexion on swing. In the P-DRFT group the patients underwent prophylactic DRFT because they had severe flexed knee gait and demonstrated normal or increased pKFSw. The purpose of this procedure was to conserve pKFSw and improve knee flexion in swing after the correction of flexed knee in stance. These two groups were evaluated separately.

Significant correlation between parameters of the left and right limbs were found ($p<0.05$). Due to this dependence, data analysis was done only for the left limbs of the patients in selected parameters from the clinical examination and 3D gait analysis. Limbs with pKFSw that was one standard deviation below the age-matched reference value (<55° of pKFSw) were assigned to the C-DRFT group (33 limbs), while the other limbs were assigned to the P-DRFT group (20 limbs) for further analysis (Fig.17).



17. Figure: Inclusion and exclusion criteria (119). Ages are given as the mean and the standard deviation. **CP**: cerebral palsy, **DRFT**: distal rectus femoris transfer, **GMFCS**: Gross Motor Function Classification System, **BTX:** botulinum toxin type A injections, **SEMLS**: single-event multilevel surgery, **C-DRFT**: group that had distal rectus femoris transfer to correct decreased peak knee flexion in swing phase, and **P-DRFT**: group that had prophylactic distal rectus femoris transfer.

For the data evaluation the following parameters in Tables 19, 20 and 21 were used. Normative data were obtained from an age-matched healthy reference group. Similar to the hamstring study, descriptive statistics were used for basic statistical analysis. For the evaluation of group differences, Levene and unpaired t tests were used. P values of <0.05 were considered significant, and Bonferroni correction was used to adjust for multiple comparisons. Person correlations were calculated between baseline parameters and their improvement between baseline and the time of the long-term follow-up. Statistical analysis was performed with use of PASW Statistics 18 (SPSS, Chicago, Illinois).

3.4. Does proximal rectus femoris release influence kinematics in patients with cerebral palsy and stiff knee gait?

All cerebral palsy patients with spastic diplegia were selected from the database, who were treated with DRFT in combination with proximal rectus femoris release. 20 patients were found who were ambulatory (GMFCS I-III) (12, 13). Their age was 11±3.4 years at surgery. All of these patients underwent the standard examinations preoperatively (E0), 1 year (E1) and at least 5 years (E2) postoperatively. As a control group other 20 patients who received only DRFT without proximal rectus femoris release were matched. The matching criteria included: maximum knee flexion in swing, knee flexion range in swing, age at surgery, BMI and GGI (59). In the group, who underwent DRFT with proximal rectus femoris release, the APT preoperatively was significantly higher, indicating that increased APT was one criterion to perform proximal rectus femoris release in this group. Further criterion was for proximal rectus femoris release a hip flexion contracture greater than 10° measured with Thomas test (109).

In the DRFT group the mean age was 9.9±2.9 years. The demographic data can be seen in the Table 9. All patients underwent SEMLS, including soft tissue and bony operations (Tabl.10). The DRFT group has received only DRFT, but the other patient group received DRFT and proximal rectus femoris release. Standard surgical techniques were used for both procedures (45, 100, 119).

9. Table: Demographic data and functional baseline parameters0 (120)

	DRFT (n=20 patients)			**DRFT with release (n=20 patients)**		
	E0	**E1**	**E2**	**E0**	**E1**	**E2**
Time of follow-up (years)	-	0.91(±0.2)	8.47(±2.1)	-	1.00(±0.1)	9.17(±2.2)
Age of patients (years)	9.94(±2.9)	11.05(±3.0)	18.31(±3.7)	11.00(±3.4)	12.29(±3.6)	20.58(±4.7)
BMI	16.83(±3.9)	17.77(±3.8)	20.66(±4.7)	18.11(±3.2)	19.29(±3.8)	21.77(±5.1)
Sex		7♀, 13♂			11♀, 9♂	
GMFCS I	2	3	5	1	3	4
GMFCS II	13	10	11	14	9	12
GMFCS III	5	7	4	5	8	4

Legend: DRFT: distal rectus femoris transfer, **GMFCS**: Gross Motor Function Classification Scale, **BMI:** Body Mass Index, **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 8 years postoperatively

10. Table: Number of surgical procedures performed in single event multilevel surgery (120)

	DRFT	**DRFT with release**
DRFT	40	40
Proximal rectus femoris release	0	40
Psoas over the brim	2	2
Hamstring lengthening	32	28
Calf muscle lengthening	32	38
Femoral derotation osteotomy distal	14	8
Femoral derotation osteotomy proximal	9	18
Tibial derotation osteotomy	3	2
Bony foot stabilization	15	12
Soft tissue foot	12	6
Total number of procedures	159	190
Average number per patient	7.95	9.5

Legend: DRFT: distal rectus femoris transfer.

For data analysis in both groups were compared using one-way repeated measures ANOVA with Tukey’s post hoc test (Prism 5; GraphPad Software, La Jolla, CA, USA) and t-test for group analysis. The level of significance was set up $p<0.05$.

3.5. Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendinous lengthening in children with spastic diplegia

The standard operation to correct flexed knee gait in spastic diplegia at the Department for Orthopaedic and Trauma Surgery, Heidelberg University Clinics was MTL surgery before 1998. Frequent occurrence of increased APT and genu recurvatum was observed. Thus, a prospective cohort study was conducted to evaluate the outcome of conversion of biarticular muscles to monoarticular muscles (CBM). In this study 25 cerebral palsy children with spastic diplegia were examined, who underwent CBM during SEMLS between 1998 and 2004. The inclusion criteria for the study were spastic diplegia, ambulatory patients (GMFCS I-III), age at surgery 6-16 years, flexed knee gait, scheduled for SEMLS. There were some exclusion criteria for the study: athetosis, previous lower limb orthopaedic surgery and BTX injection less than 6 month before surgery. The children underwent standard clinical examinations and 3D gait analysis before and 1 year after surgery. The results were published in 2004 (34).

For a long-term follow-up the same patients were invited at least 6 years after their multilevel surgery for the present study. Only 21 children of the 25 could be reevaluated 6-14 years after surgery. 2 patients have moved, 1 patient was not able to attend and 1 patient preferred not to participate in this study. For CBM patients 21 diplegic patients with flexed knee gait from the database were matched who have had MTL during SEMLS. The matching criteria were the following preoperative parameters: knee flexion and ankle dorsiflexion in stance (primary matching parameters), together with pelvic tilt, hip flexion, age at surgery, body mass index (BMI), Gillette Gait Index (GGI), and GMFCS level (secondary matching parameters) (59). Between the 2 groups there were no significant differences in matched preoperative parameters (one-way ANOVA, $p < 0.05$).

The reevaluated 42 patients (21 CBM, 21 MTL) were measured before the orthopaedic surgery (E0), 1 year (E1) and 6-14 years postoperatively. The demographic data's, global walking ability and surgical procedures were summarized in Table 11 and Table 12.

11. Table: Demographic data and functional baseline parameters (10)

Parameters	CBM			MTL		
	E0	E1	E2	E0	E1	E2
Gender	8♀, 13♂	8♀, 13♂	8♀, 13♂	6♀, 15♂	6♀, 15♂	6♀, 15♂
Time of follow-up (years)	-	1.3 (±0.6)	9.2 (±2.5)	-	1.2 (±0.7)	9.1 (±2.6)
Age of patients (years)	11.3 (±3.1)	12.6 (±3.2)	20.5 (±4.3)	11.1 (±5.4)	12.3 (±3.7)	20.2 (±4.5)
BMI (kg/m2)	18.3 (±3.7)	19.4 (±3.6)	20.9 (±4.0)	17.3 (±3.1)	18.1 (±3.1)	21.4 (±4.8)
GMFCS I	3	6	4	3	7	5
GMFCS II	11	5	9	13	6	10
GMFCS III	7	10	8	5	8	6
GGI	407 (295) #+	227 (160) ×+	270 (163) ×#	364 (175) #+	273 (221) ×	233 (121) ×

Legend: CBM: conversion of biarticular muscles to monoarticular muscles, **MTL:** muscle tendon lengthening surgery

BMI: Body Mass Index, **GMFCS:** Gross Motor Function Classification Scale, **GGI:** Gillette Gait Index, **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 9 years postoperatively

ANOVA was used for statistical analysis. Bonferroni corrected; level of significance $p<0.05$;

$^{\times}$significant difference to pre-operative,

$^{\#}$ significant difference to 1 year postoperative,

$^{+}$significant difference to 9 year postoperative.

All patients underwent standardized SEMLS (Tabl.12). All procedures were performed according to specific clinical and gait analysis criteria. There was a difference in the treatment principles between the two groups (CBM and MTL) concerning the following biarticular muscles: semitendinosus, gastrocnemius and rectus femoris muscle. In the CBM group, with the patient prone, a midline longitudinal incision was made in the popliteal fossa. The medial tendon origin of gastrocnemius was released at the femoral condyle, leaving a stump of 3-4 cm. The lateral head of gastrocnemius was also released, and both heads were transferred and sutured to the tibial condyles in order to convert the gastrocnemius into a monoarticular muscle. The released distal tendon of the semitendinosus was sutured to the medial gastrocnemius tendon stump. The proximal part of rectus femoris was released at the anterior iliac spine capsule in all CBM patients and sutured to the anterior hip joint capsule, while the reflected head of the rectus femoris muscle was released.

In the MTL group the semitendinosus lengthening occurred intramuscular in the middle thigh, supine position or in prone position popliteal a Z-lengthening. The gastrocnemius was lengthened by intramuscular aponeurotic lengthening. In some cases proximal rectus femoris release was performed, when the patients had a double-bump pattern of the pelvis and a positive Duncan-Ely test in the MTL group.

12. Table: Number of surgical procedures performed in single event multilevel surgery (10)

Surgical procedures	CBM	MTL
Intramuscular psoas lengthening	7	6
Adductor longus recession	6	10
Proximal rectus femoris release	42	18
Distal rectus femoris transfer	42	38
Semitendinosus transfer to distal femur	42	0
Intramuscular semitendinosus tenotomy	0	38
Semitendinosus Z-lengthening	0	4
Semimembranosus aponeurotic recession	42	42
Lateral hamstring lengthening	6	9
Gastrocnemius transfer to proximal tibia	42	0
Gastroc/soleus recession	12	29
Tendon Achilles lengthening	4	4
Soft tissue foot	9	9
Femoral derotation osteotomy (proximal)	17	18
Femoral derotation osteotomy (distal)	0	10
Tibial internal rotation osteotomy	5	5
Bony foot correction	9	15

Legend: CBM: conversion of biarticular muscles to monoarticular muscles, **MTL:** muscle tendon lengthening surgery

Postsurgical for the first days epidural anesthesia was used to decrease postoperative pain and early mobilization with passive ROM treatment was started. For 4 weeks all of the patients were lower leg weight-bearing casts and altogether for 6 months a long-leg night orthoses to maintain knee extension. After 4 weeks ankle-foot orthoses with dorsiflexion stop were fitted to assist passive extension of the knee. In those patients, who have had bony procedures, short-leg non-weight-bearing casts were fitted for 4 weeks after operation.

Basic descriptive statistical analysis was done using PASW® Statistics 18 and were used both limbs of each patient. The GGI was calculated for all subjects at all examinations. Two-way repeated-measures analyses of variance (ANOVA) were applied to show time and group effects. Significant difference was accepted, if p-values<0.05 and Bonferroni correction was employed to adjust multiple comparisons.

4. Results

4.1. Development of knee function after hamstring lengthening as a part of multilevel surgery in children with spastic diplegia

4.1.1. Clinical examinations

A significant reduction of mean popliteal angle could be found at E1 for the MHL and the CHL group (Tabl.13, Fig.18). Between E1 and E3, the two groups showed significant differences in the development of popliteal angle. The popliteal angle deteriorated mainly between E2 and E3 in both groups. In long-term follow-up examination, the popliteal angle was not significant different from the pre-operative values in either groups.

13. Table: The change of popliteal angle (9)

Parameters	E0	E1	E2	E3
Popliteal angle (°)				
All	51 (±18)#**	30 (±19)§††	38 (±16)§††	49 (±16)#**
MHL	47 (±17)#	28 (±19) §††	37 (±15)††	51 (±15)#**
CHL	66 (±11)#	38 (±17) §	43 (±19)	43 (±21)

Legend: MHL: medial hamstring lengthening, **CHL**: combined medial and lateral hamstring lengthening, **E1**: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively.
Significance was determined with repeated measures of analysis of variance with Bonferroni correction. The level of significance was set at $p<0.05$
§Significantly different from E0.
#Significantly different from E1.
**Significantly different from E2.
††Significantly different from E3.

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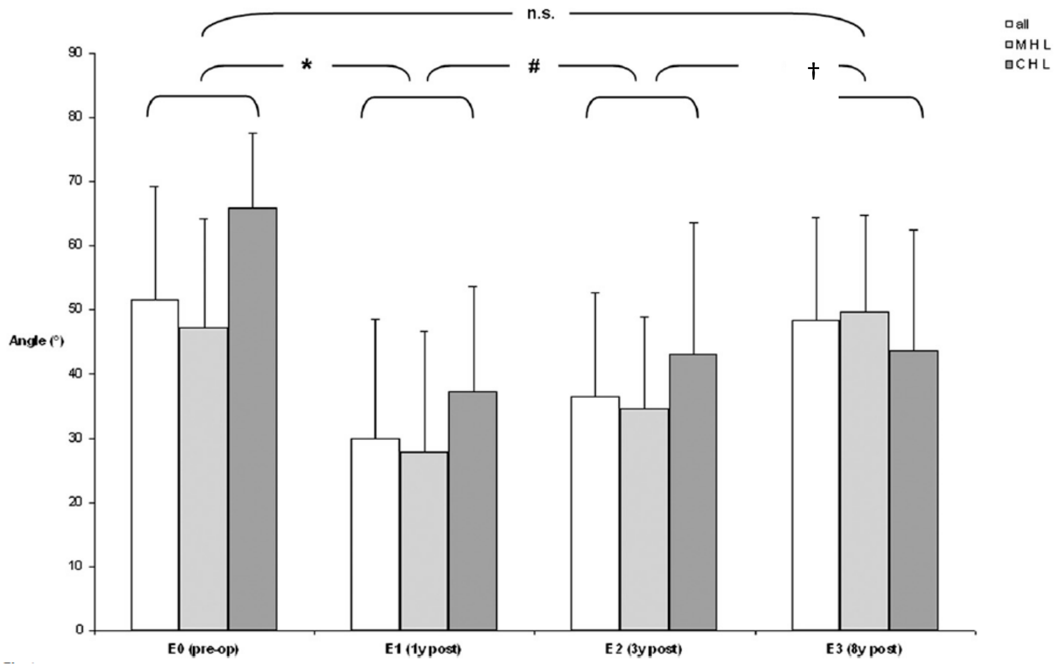


18. Figure: Histogram showing the development of the popliteal angle (given as the mean and the standard deviation) at E0 (preoperatively), E1 (1 year postoperatively), E2 (2-4 years postoperatively), and E3 (6-12 years postoperatively) for all patients (n = 39), for the medial hamstring lengthening (MHL) group (n= 30 patients), and the combined medial and lateral hamstring lengthening (CHL) group (n= 9 patients). Positive values indicate knee flexion.*Significant difference between E0 and E1 ($p < 0.001$). #Significant difference between E1 and E2 ($p < 0.01$). †Significant difference between E2 and E3 ($p < 0.01$). n.s. = not significant.(9)

4.1.2. Global gait variables

The GMFCS levels have improved over the years, 7 more patients showed GMFCS I at E3 in comparison with E0. But there was no significant difference in the GMFCS changes between the 2 groups: MHL and CHL.

The GGI was found reduced one year after surgery in both groups. The improvements of GGI were maintained in the long-term follow-up (Tabl.14).

14. Table: The change of GGI (9)

Parameters	E0	E1	E2	E3
GGI				
All	378 (±240)#**††	225 (±172)§	207 (±168)§	217 (±134)§
MHL	377 (±267) #**††	207 (±142)§	201 (±177)§	208 (±130)§
CHL	380 (±129)**	287 (±249)	228 (±139)§	246 (±149)

Legend: MHL: medial hamstring lengthening, **CHL**: combined medial and lateral hamstring lengthening, **E1**: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively.
Significance was determined with repeated measures of analysis of variance with Bonferroni correction. The level of significance was set at p<0.05.
§Significantly different from E0.
#Significantly different from E1.
**Significantly different from E2.
††Significantly different from E3.

4.1.3. 3D gait analysis

4.1.3.1. Temporal parameters

Patients of both groups walked slower initially after surgery but increased walking speed was seen from E1 to E3 for the MHL group. Cadence initially decreased after surgery, but increased in the following years equally in both groups. Stride length showed a significant increase between E1 and E3 for the patients of the MHL group. The differences in the development of time-distance parameters between both groups showed no significance (Tabl.15).

15. Table: The change of temporal parameters (9)

Parameters	E0	E1	E2	E3
Speed (m/s)				
All	0.82 (±0.23)#	0.70 (±0.32)§**††	0.87 (±0.30)#	0.92 (±0.30)#
MHL	0.83 (±0.23)#††	0.70 (±0.30) §**††	0.88 (±0.28)	0.96 (±0.28)#
CHL	0.78 (±0.21)	0.71 (±0.41)	0.84 (±0.37)	0.79 (±0.35)
Cadence (steps/min)				
All	120 (±24)#	100 (±30)§**††	113 (±26)	119 (±21)
MHL	123 (±26)#	104 (±27)§	115 (±25)	114 (±20)
CHL	113 (±13)	88 (±37)	104 (±27)	104 (±23)
Stride length (m)				
All	0.82 (±0.19)**††	0.82 (±0.24)**††	0.91 (±0.23)§#	0.97 (±0.21)§#
MHL	0.82 (±0.21)††	0.79 (±0.23) **††	0.90 (±0.23)#††	0.99 (±0.20)§#**
CHL	0.82 (±0.12)	0.91 (±0.23)	0.95 (±0.26)	0.89 (±0.24)

Legend: MHL: medial hamstring lengthening, **CHL**: combined medial and lateral hamstring lengthening. **E1**: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively.
Significance was determined with repeated measures of analysis of variance with Bonferroni correction. The level of significance was set at p<0.05.
§Significantly different from E0.
#Significantly different from E1.
**Significantly different from E2.
††Significantly different from E3.

4.1.3.2. Kinematics

An increased mean pelvic tilt was found 1 year after surgery for both groups, especially for the CHL group (Tabl.16). In the mid-term follow-up the pelvic tilt significantly decreased in both groups, whereas an increase was noted in the long-term follow-up.

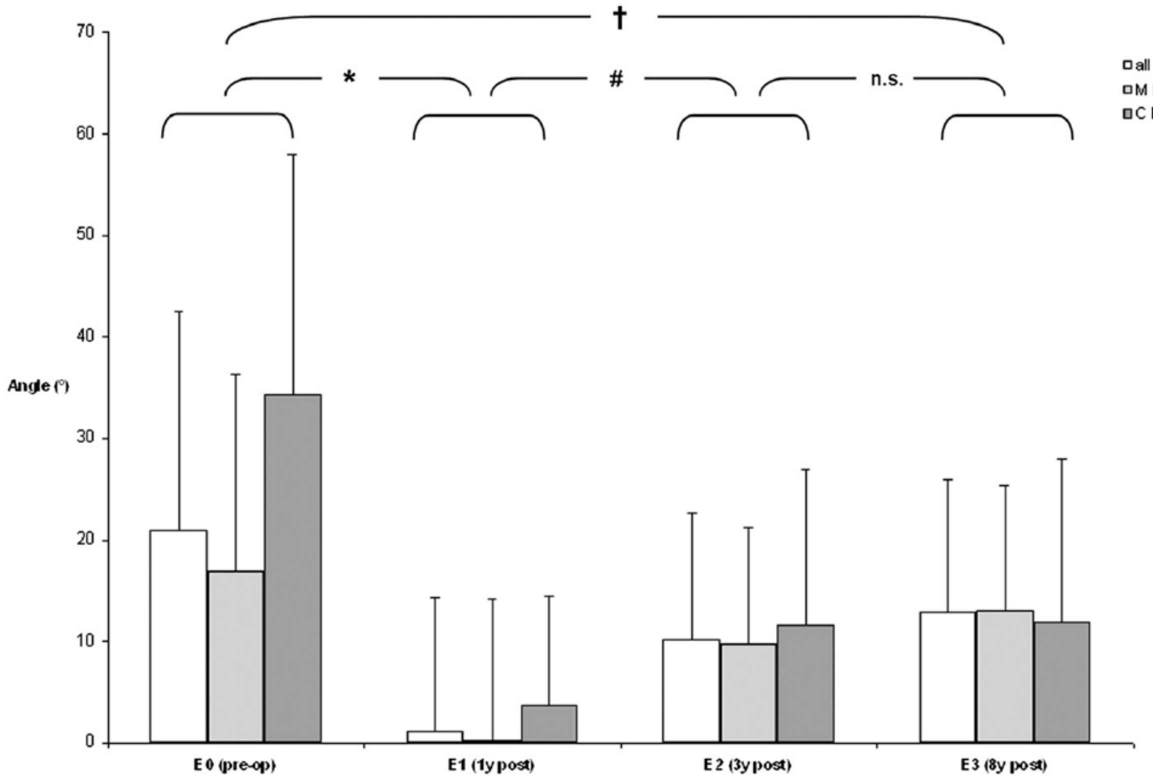
Concerning knee kinematics, the CHL group showed a higher extent of flexed knee gait, represented by the parameters “knee flexion at initial contact”, “minimum knee flexion in stance” and “mean knee flexion in stance” in comparison to the MHL group pre-operatively. A significant reduction of all the three knee kinematic parameters could be found in both groups initially after surgery (Tabl.16). Minimum knee flexion in stance was significantly reduced by 17° for the MHL and 28° for the CHL group to reference values. Over the years, a significant recurrence was found in all three knee kinematic parameters for the whole group of patients (Tabl.16, Fig.19), with 9° of deterioration noted for minimum knee flexion in stance. The majority of improvements could be seen in the first 3 years after surgery. While the MHL group showed significant recurrence of minimum knee flexion in stance, the slight deterioration in CHL group was not significant. Comparing long-term outcome (±E3) and baseline (E0) of both groups, the MHL group did not show significant differences whereas in the CHL group significant differences could be found.

For illustration, average (and SD) sagittal plane kinematics at baseline, at E1, at E2 and at E3 are shown in Figure 20. Additionally, reference data of physiologic gait are indicated (24 age-matched normalized patients). Beside pelvic and knee kinematics, hip and ankle kinematics are also shown because crouch gait is influenced by hip and ankle position.

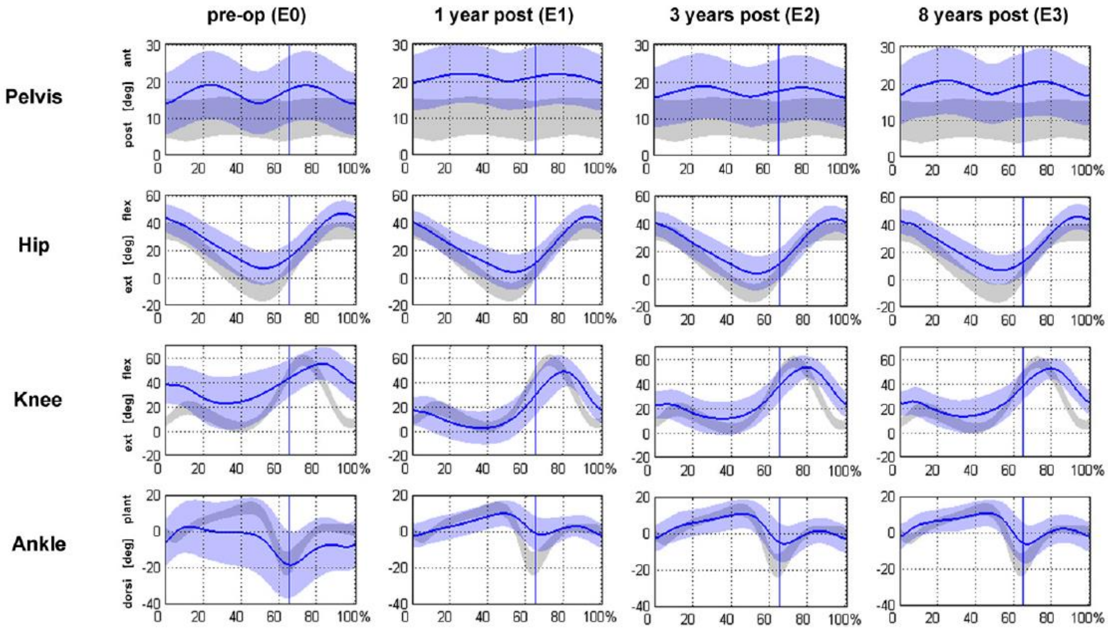
16. Table: The change of kinematic parameters (9)

Parameters	E0	E1	E2	E3
Mean pelvic tilt (°)				
All	17 (±9)	21 (±8)**	17 (±8)#	18 (±8)
MHL	17 (±8)	20 (±7) **	17 (±8)#	18 (±7)
CHL	16 (±10)	22 (±10)	17 (±10)	20 (±10)
Knee flexion at initial contact (°)				
All	38 (±17)#**††	17 (±11)§**††	22 (±11)§#	23 (±10)§#
MHL	36 (±17) #**††	16 (±12)§**††	22 (±11)§#	23 (±10)§#
CHL	45 (±14) #**††	19 (±8)§	20 (±12)§	23 (±10)§
Mean knee flexion in stance (°)				
All	30 (±19) #**††	11 (±12)§**††	18 (±13)§#	20 (±11)§#
MHL	27 (±18) #**	10 (±13)§**††	17 (±12)§#	20 (±11)#
CHL	42 (±20) #††	15 (±12)§	20 (±16)	21 (±14)§
Minimum knee flexion in stance (°)				
All	21 (±22) #**††	1 (±14)§**††	10 (±13)§#	12 (±13)§#
MHL	17 (±20) #**	0 (±14)§**††	9 (±12)§#	12 (±12)#
CHL	35 (±23) #††	7 (±12)§	12 (±16)	12 (±16)§

Legend: MHL: medial hamstring lengthening, **CHL**: combined medial and lateral hamstring lengthening, **E1**: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively.
Significance was determined with repeated measures of analysis of variance with Bonferroni correction. The level of significance was set at $p<0.05$.
§Significantly different from E0.
#Significantly different from E1.
**Significantly different from E2.
††Significantly different from E3.



19. Figure: Histogram showing the development of minimum knee flexion in stance (given as the mean and the standard deviation) at E0 (preoperatively), E1 (one year postoperatively), E2 (two to four years [mean, three years] postoperatively), and E3 (six to twelve years [mean, eight years] postoperatively) for all patients (n = 39), for the medial hamstring lengthening (MHL) group (n = 30), and for the combined medial and lateral hamstring lengthening (CHL) group (n = 9). Positive values indicate knee flexion. *Significant difference between E0 and E1 ($p < 0.001$). Significant difference between E0 and E3 ($p < 0.01$). #Significant difference between E1 and E2 ($p < 0.05$). n.s. = not significant (9).



20. Figure: Average sagittal plane kinematics as shown on gait analysis for all patients (n = 39) at E0 (preoperatively), E1 (one year postoperatively), E2 (two to four years [mean, three years] postoperatively), and E3 (six to twelve years [mean, eight years] postoperatively). The mean is represented by the blue line, the standard deviation is represented by the lucent blue area, and age-matched normalized data are represented by the gray area. Positive values indicate anterior pelvic tilt, hip flexion, knee flexion, and ankle dorsiflexion (9).

4.1.3.3. Kinetics

Kinetic data could not be recorded for patients using a walking device. In 19 out of 39 patients kinetic data was enclosed in this study due to the fact that those patients were able to walk free at all four examination dates. The maximum internal knee flexion moment was found significantly reduced at E1 for these 19 patients (Tabl.17). There was a significant recovery of knee flexor moment between E1 and E2, which was maintained at E3, but pre-operative values were not reached again. The maximum knee extensor moment increased over the years. These changes were not significant.

17. Table: The change of kinetic parameters (9)

Parameters	E0	E1	E2	E3
Maximum internal knee flexor moment (Nm/kg*m)	0.55 (±0.27) #	0.40 (±0.21) §**	0.52 (±0.27) #	0.55 (±0.24)
Maximum internal knee extensor moment (Nm/kg*m)	-0.37 (±0.25)	-0.47 (±0.29)	-0.42 (±0.22)	0.50 (±0.31)

Legend: E1: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively.
Significance was determined with repeated measures of analysis of variance with Bonferroni correction. The level of significance was set at $p<0.05$.
§Significantly different from E0.
#Significantly different from E1.
**Significantly different from E2.

4.1.4. Adverse effects

The number of limbs showing knee recurvation was calculated (Tabl.18). Genu recurvatum was defined as knee hyperextension of more than 5° in stance phase. One year postoperatively a recurvatum knee was found in 18 patients (35%, bilateral involvement in 9 patients and unilateral involvement in 9 patients). In this recurvation group the number decreased significantly between E1 and E2 to 10 legs (13%, 6 limbs bilateral and 4 limbs unilateral), which was maintained at the long-term follow-up in 9 limbs (12%). There were no cases developing recurvation after E1. No significant differences in occurrence of genu recurvatum were found comparing the groups MHL and CHL in any of the postoperative examinations. At E1, genu recurvatum occurred mainly in Groups II and III (representing 56% and 26% of the cases, respectively), with only 19% of the cases occurring in Group IV (the crouch group). At E3 there were no knee hyperextensions in Group IV, whereas residual genu recurvatum was observed in nine limbs in Groups II (8) and III (1).

A significant increase of mean pelvic tilt was found in 38 legs (49%) 1 year after surgery (Tabl.18). Patients who underwent combined medial and lateral hamstring lengthening (CHL) showed increased pelvic tilt in 67% (12 of 18 legs) in comparison to the isolated medial hamstring group (MHL) where in 43% (26 of 60 legs) increased pelvic tilt was found.

The incidence of increased pelvic tilt number decreased slightly between E1 and E2 to 30 legs (38%) but increased in the long-term follow-up to 46% (36 of 78 legs).

18. Table: Adverse effects (9)

	E1	E2	E3
Genu recurvatum ‡			
All (n=78)	27	10§	9§
MHL (n=60)	21	7	7
CHL (n=18)	6	3	2
Group II (jump knee) (n=33)#	15	8	8
Group III (apparent equinus) (n=33) #	7	2	1
Group IV (crouch) (n=12) #	5	0	0
Increased pelvic tilt *			
All (n=78)	38	30§	36§
MHL (n=60)	26	22	24
CHL (n=18)	12	8	12

Legend: The total number of limbs showing genu recurvatum or increased pelvic tilt is shown for all groups and examinations. **MHL**: medial hamstring lengthening, **CHL**: combined medial and lateral hamstring lengthening, **E1**: 1 year postoperatively, **E2**: 2-4 years postoperatively, **E3**: 6-12 years postoperatively.
‡Genu recurvatum is defined as >5° of extension in stance phase.
§Significantly different from the previous time period (McNemar test).
#Sagittal gait pattern classification according to Rodda et al. obtained from preoperative, instrumented, 3D gait analysis (80).
*Increased pelvic tilt is defined as an increase of mean pelvic tilt of >5° in comparison with E0.

4.1.5. Correlation between static and dynamic parameters

The correlation coefficient between the deterioration of popliteal angle (difference between E1 and E3) and the recurrence of flexed knee gait on gait analysis, represented by minimum knee flexion in stance, was calculated for all legs. Only a poor correlation between the static (popliteal angle) and the dynamic parameter (minimum knee flexion in stance) was found (rp=0.29; p=0.09; Pearson correlation).

4.2. Long-term results after distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff knee gait in spastic diplegic cerebral palsy

4.2.1. Clinical examination

Significantly less patients showed a positive Duncan-Ely sign one year after DRFT ($p<0.01$), especially in the C-DRFT (DRFT for correction of stiff knee gait) group (Tabl.19). However, between E1 and E2 examination the number of patients with positive Duncan-Ely sign increased significantly in both groups. The increase of Duncan-Ely sign was significantly higher in the P-DRFT group ($p<0.05$), and no significant difference was found between E0 and E2. In the C-DRFT group 6 limbs showed moderate knee flexion contracture between 10° and 20° preoperatively. In the P-DRFT group, 6 limbs had moderate and 10 had severe (between 20° and 40°) knee flexion contracture preoperatively.

19. Table: Outcomes of clinical parameters (119)

Parameters	E0	E1	E2	Group difference
Duncan-Ely sign. (no. of patients)				
C-DRFT	33§#	12#**	21§**	E1, E2
P-DRFT	20§#	12#**	18§**	
Passive knee extension (°)				
C-DRFT	-3 (±6)§#	1 (±7)**	2 (±6)**	E0, E2
P-DRFT	-13 (±11)§#	-2 (±7)**	-2 (±8)**	
Passive knee flexion (°)				
C-DRFT	138 (±15)	139 (±12)	142 (±6)	-
P-DRFT	136 (±18)	139 (±11)	133 (±16)	

Legend: The Levene and unpaired t tests were used to detect differences between groups E0, E1 and E2, and Bonferroni correction was used. The level of significance was a p value of <0.05. **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 8-10 years postoperatively. **C-DRFT**: DRFT for correction of stiff knee gait, **P-DRFT**: prophylactic DRFT by flexed knee gait
§ Significant difference compared with E1.
# Significant difference compared with E2.
** Significant difference compared with E0.

4.2.2. Global gait variables

The Gillette Gait Index (GGI) as a global walking function parameter was significantly reduced one year after surgery and maintained in the long-term follow-up in both groups (Tabl.20). The patients in the P-DRFT group had severe flexed knee contracture. Hence, their GGI level was significantly higher preoperatively ($p<0.01$), and the overall improvement was smaller in comparison with that of the C-DRFT (DRFT for correction of stiff knee gait) group.

Altogether the GMFCS level was significantly decreased and this was maintained in the long-term follow-up examination in both groups. In the C-DRFT group 1 year after surgery 3 patients with GMFCS II improved to level I and 4 patients with GMFCS II needed walking devices (level III). At E2 two patients with GMFCS II preoperatively reached level I. By 2 patients, who had preoperatively GMFCS III improved to level II at E2. Two other patients with level II preoperatively needed walking devices at E2.

In the P-DRFT group at the long-term follow-up, 2 patients with GMFCS II reached level I, 4 patients with GMFCS III reached level II and 2 patients with GMFCS II were classified as level III.

20. Table: The change of GGI (119)

Parameters	**E0**	**E1**	**E2**	**Group difference**
GGI				
C-DRFT	361 (±347)§#	136 (±105)**	160 (±121)**	E1, E2
P-DRFT	486 (±282)#	319 (±198)	290 (±176)**	

Legend: The Levene and unpaired t tests were used to detect differences between groups E0, E1 and E2, and Bonferroni correction was used. The level of significance was a p value of <0.05. **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 8-10 years postoperatively. **C-DRFT**: DRFT for correction of stiff knee gait, **P-DRFT**: prophylactic DRFT by flexed knee gait
§ Significant difference compared with E1.
# Significant difference compared with E2.
** Significant difference compared with E0.

4.2.3. 3D gait analysis

4.2.3.1. Temporal parameters

Step length did not show a significant increase between preoperative and one year follow-up examination in either group, but increased significantly between E1 and E2 in C-DRFT group ($p<0.001$). This increase may be explained by the increase of the leg length during growth. Walking speed initially was significantly decreased in the P-DRFT group but increased between E1 and E2. For the C-DRFT group a significant increase in walking speed was found between E0 and E2. Cadence was not significantly increased comparing preoperative and long-term follow-up examinations (Tabl.21).

21. Table: Outcomes of temporal parameters (119)

Parameters	**E0**	**E1**	**E2**	**Group difference**
Timing of toe-off (% GC)				
C-DRFT	65 (±5)	65 (±4)	64 (±4)	E2
P-DRFT	67 (±5)§	71 (±9)**	68 (±7)	
Step length (m)				
C-DRFT	0.4 (±0.1)#	0.4 (±0.1)#	0.5 (±0.1)§**	E2
P-DRFT	0.4 (±0.1)	0.4 (±0.1)	0.4 (±0.1)	
Speed (m/s)				
C-DRFT	0.9 (±0.3)#	0.9 (±0.2)#	1.0 (±0.3)§	E0, E1, E2
P-DRFT	0.7 (±0.2)	0.6 (±0.3)	0.7 (±0.3)	
Cadence (steps/min)				
C-DRFT	125 (±24)§**	115 (±19)	114 (±21)	E0, E1, E2
P-DRFT	110 (±26)§	82 (±33)**	96 (±24)	

Legend: The Levene and unpaired t tests were used to ditect differences between groups E0, E1 and E2, and Bonferroni correction was used. The level of significance was a p value of <0.05. **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 8-10 years postoperatively. **C-DRFT**: DRFT for correction of stiff knee gait, **P-DRFT**: prophylactic DRFT by flexed knee gait
§ Significant difference compared with E1.
# Significant difference compared with E2.
** Significant difference compared with E0.

4.2.3.2. Kinematics

The range of knee motion (ROM) during swing phase was significantly increased one year after surgery in both groups ($p<0.001$), but this increase was significantly higher in the C-DRFT group ($p<0.01$). This increase was maintained in the long-term follow-up in the C-DRFT group, whereas a decrease ($p=008$) was found in the P-DRFT.

Concerning peak knee flexion in swing phase, an increase was found 1 year after DRFT in C-DRFT group which showed statistical tendency. Between E1 and E2 another increase was found. Comparing the peak knee flexion preoperatively with the long-term outcome value, a significant and maintained increase was found in C-DRFT group ($p<0.01$). In the P-DRFT group there was found first a significant decrease 1 year postoperatively and at E2 the peak knee flexion in swing tended to increase, but this increase remained significantly decreased in comparison with the preoperatively value (from 67° to 54°) (Tabl.22).

Timing of peak knee flexion in swing was found to be significantly reduced ($p<0.01$) after DRFT in C-DRFT at one year follow-up examination and this reduction was maintained at E2 examination ($p<0.01$), whereas no significant changes was detected for the P-DRFT group.

The stance phase parameters (mean knee flexion in stance, knee flexion at initial contact and minimum knee flexion in stance) were significantly decreased from baseline in both groups, although this decrease was significantly higher in the P-DRFT group because of the significantly greater knee flexion contracture at baseline in comparison with the C-DRFT group. Between E1 and E2 a deterioration of stance phase parameters was detected in both groups, but this deterioration was significantly higher in the P-DRFT group ($p<0.01$).

A significant increase in knee flexion velocity was found after DRFT in both groups ($p<0.01$). At long-term follow-up this increase was maintained and the difference between E2 and E0 was still significant in both groups ($p<0.01$).

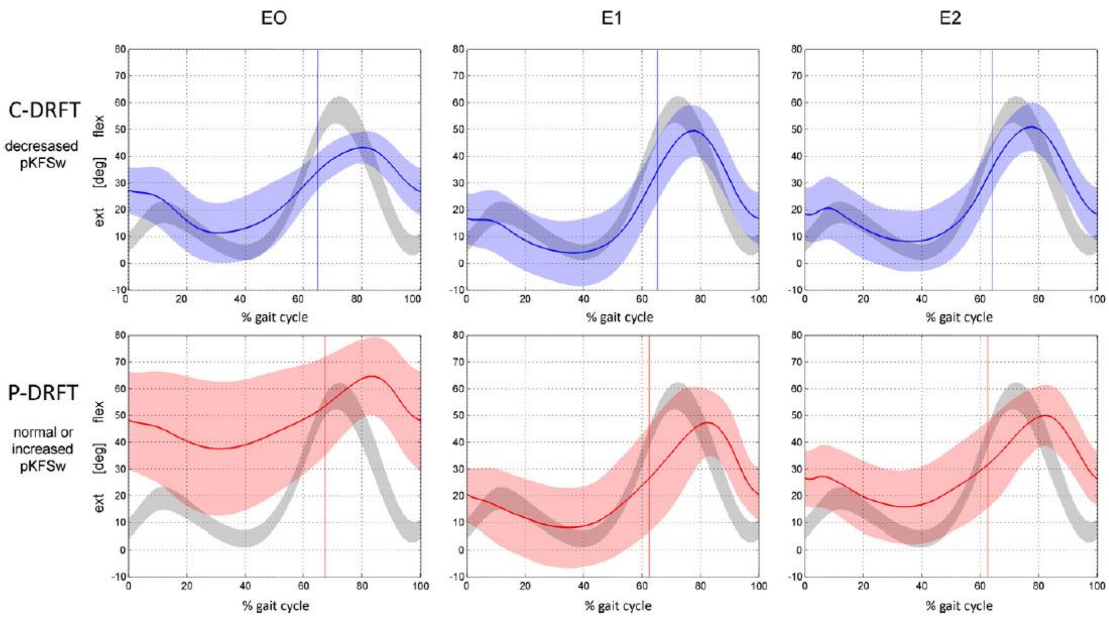
The mean knee kinematic curves of patients and norm at all three examinations are visualized in Figure 21.

22. Table: Outcomes of kinematics parameters (119)

DOI:10.14753/SE.2016.1896

Parameters	E0	E1	E2	Group difference
Range of knee flexion in gait cycle (°)				
C-DRFT	35 (±11)§#	49 (±12)**	47 (±11)**	E2
P-DRFT	29 (±15)§#	45 (±14)**	40 (±13)**	
Range of knee flexion in stance (°)				
C-DRFT	26 (±9)§	33 (±10)#**	29 (±8)§	E0
P-DRFT	19 (±9)§#	34 (±12)#**	26 (±10)§**	
Range of knee flexion in swing (°)				
C-DRFT	20 (±8)§#	35 (±10)**	34 (±10)**	E2
P-DRFT	19 (±8)§#	32 (±10)**	29 (±9)**	
Knee flexion in stance (°)				
C-DRFT	19 (±10)§	11 (±10)**	14 (±10)	E0, E2
P-DRFT	45 (±18)§#	16 (±14)**	23 (±12)**	
Knee flexion at initial contact (°)				
C-DRFT	27 (±9)§#	17 (±9)**	18 (±9)**	E0, E2
P-DRFT	49 (±17)§#	21 (±10)#**	27 (±9)§**	
Minimum knee flexion in stance (°)				
C-DRFT	10 (±11)§	2 (±12)**	5 (±9)	E0, E2
P-DRFT	38 (±20)§#	7 (±14)#**	15 (±13)§**	
Peak knee flexion in swing (°)				
C-DRFT	45 (±6)§#	51 (±9)**	52 (±9)**	E0
P-DRFT	67 (±12)§#	52 (±12)**	54 (±7)**	
Timing of peak knee flexion in swing (% GC)				
C-DRFT	80 (±6)§#	77 (±4)**	77 (±4)**	E1, E2
P-DRFT	82 (±5)	81 (±6)	81 (±5)	
Knee flexion velocity (°/% GC)				
C-DRFT	0.8 (±0.3)§#	1.2 (±0.4)**	1.1 (±0.4)**	E2
P-DRFT	0.6 (±0.3)§#	1.0 (±0.4)**	0.9 (±0.4)**	

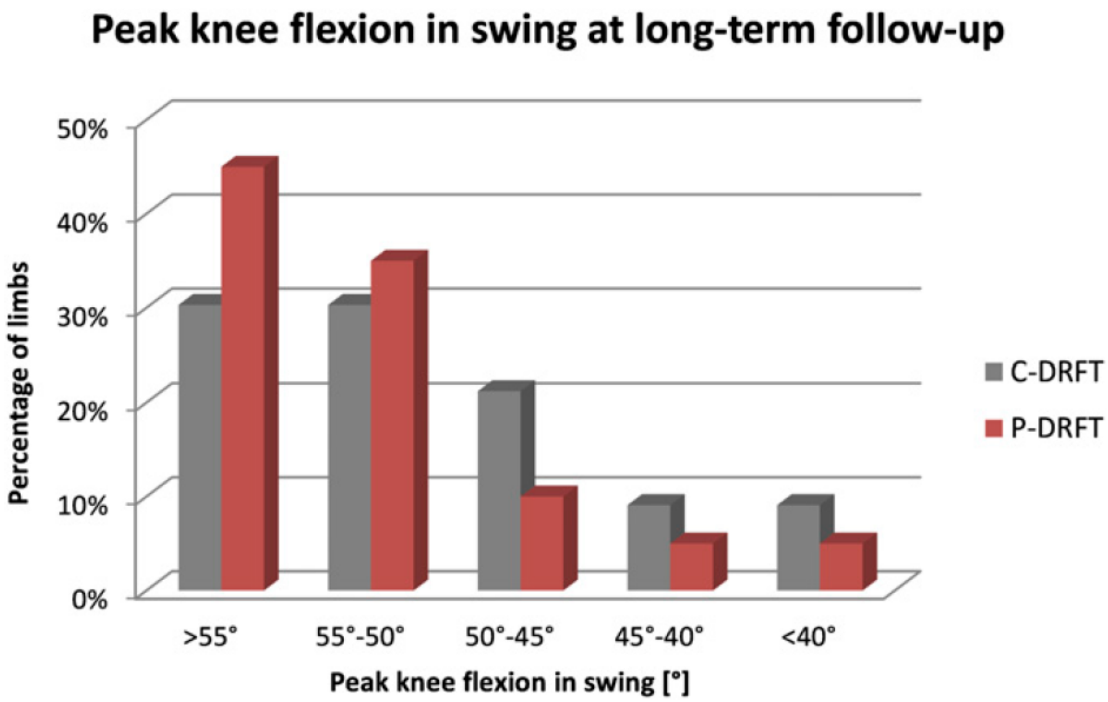
Legend: The Levene and unpaired t tests were used to detect differences between groups E0, E1 and E2, and Bonferroni correction was used. The level of significance was a p value of <0.05. **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 8-10 years postoperatively. **C-DRFT**: DRFT for correction of stiff knee gait, **P-DRFT**: prophylactic DRFT by flexed knee gait. § Significant difference compared with E1. # Significant difference compared with E2. ** Significant difference compared with E0.



21. Figure: Sagittal plane knee kinematics (119). Graphs are shown for the preoperative (E0; left column), one-year postoperative (E1; middle column), and nine-year postoperative (E2; right column) examinations. In the top row, the mean patient data for the C-DRFT group are represented by the blue line and the standard deviation is displayed as the lucent blue area. In the bottom row, the mean patient data for the P-DRFT group are represented by the red line and the standard deviation is displayed as the lucent red area. The reference data for physiologic knee kinematics obtained by an age-matched group of twenty-five normal subjects are also presented (gray area). Positive values indicate knee flexion. pKFSw = peak knee flexion in swing. C-DRFT: DRFT for correction of stiff knee gait, P-DRFT: prophylactic DRFT by flexed knee gait

4.2.4. Response and recurrence

With regard to knee flexion in swing, a good response was defined as an increase of knee flexion in swing of at least 10º at one year postoperatively. Accordingly, an increase of <10º was classified as a poor response. At one year, twenty-two limbs (67%) in the C-DRFT group and fifteen limbs (75%) in the P-DRFT group were classified as having a good response. Eleven limbs (33%) in the C-DRFT group and five limbs (25%) in the P-DRFT group showed a poor response. At long-term follow-up, the groups showed comparable rates of limbs with a good response (67% in the C-DRFT group and 65% in the P-DRFT group), whereas 15% and 5%, respectively, had a late response. Eighteen percent of the C-DRFT group and 15% of the P-DRFT group showed a permanently poor response. The preoperative and nine-year postoperative knee kinematics of a typical patient with a poor response are illustrated in the Appendix. Fifteen percent of the C-DRFT group and 20% of the P-DRFT group developed recurrence of a stiff-knee gait with regard to knee flexion in swing. With regard to peak knee flexion in swing, 39% of the limbs in the C-DRFT group and 20% of the limbs in the P-DRFT group had peak knee flexion in swing below two standard deviations (<50°) of the age-matched reference value at the time of the long-term follow-up (Fig.22). Distal rectus femoris transfer was not able to increase (C-DRFT group) or to maintain (P-DRFT group) peak knee flexion in swing in those patients.



22. Figure: The percentage of limbs in different categories of resulting peak knee flexion in swing at the time of the long-term follow-up in the group that had distal rectus femoris transfer for correction of decreased peak knee flexion (C-DRFT; gray bars) and the group with normal or increased peak knee flexion that had prophylactic distal rectus femoris transfer (P-DRFT; red bars). Thirty-nine percent of the limbs in the C-DRFT group and 20% of the limbs in the P-DRFT group presented with peak knee flexion in swing below two standard deviations (<50º) of the age-matched reference value at the time of the long-term follow-up (119). C-DRFT: DRFT for correction of stiff knee gait, P-DRFT: prophylactic DRFT by flexed knee gait

4.2.5. Correlations

A significant correlation (r_p = -0.8; $p < 0.0001$) was found between the peak knee flexion in swing and the amount of its improvement (the difference between preoperative and long-term follow-up evaluations). The patients with more involvement have more potential to benefit from distal rectus femoris transfer. In patients with >53º of peak knee flexion in swing preoperatively, an increase of peak knee flexion in swing is not to be expected.

4.3. Does proximal rectus femoris release influence kinematics in patients with cerebral palsy and stiff knee gait?

4.3.1. Global gait variables

The GMFCS levels have improved over the years. Altogether in two groups 6 more patients showed GMFCS I at E2 in comparison with E0: 3 more patients in DRFT group and 3 more patients in DRFT with release group. There was no significant difference in the GMFCS changes between the 2 groups: DRFT and DRFT with release (Tabl.9).

The GGI reduced significantly one year after surgery in both groups and these improvements were maintained in the long-term follow-up (Tabl.23). Between the 2 groups no significant differences were observed.

23. Table: The change of GGI (120)

	E0	**E1**	**E2**
GGI			
DRFT	426 (±541)	212 (±194)*	193 (±116)*
DRFT with release	350 (±240)	187 (±160)*	199 (±198)*

Legend: GGI: Gillette Gait Index, **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 8 years postoperatively

4.3.2. 3D gait analysis

4.3.2.1. Temporal parameters

The normalized cadence showed no significant decrease in both groups at E1 without any group differences. At E2 the normalized cadence tended to increase in both groups. This improvement was higher in the DRFT with release group, but there was no significant difference between 2 groups. 1 year postoperatively the normalized speed decreased slightly in both groups without any group differences. At E2 it was increased in both groups, whereas this improvement was significant only in DRFT group. At long-term follow-up the normalized stride length improved in both groups, but this change was significant only in the DRFT group. There was no significant group difference in normalized stride length between the 2 groups at E2 (Tabl.24).

24. Table: Outcomes of temporal parameters (120)

	Normal reference	E0	E1	E2
Timing of toe-off (%GC)	60 (±1)			
DRFT		66 (±5)	68 (±6)	64 (±4)*#
DRFT with release		64 (±5)	67 (±6)*	64 (±6)#
Normalized cadence (steps/min)†	-			
DRFT		43.4(±7.5)	40.0(±9.1)	44.4(±6.6)
DRFT with release		44.7(±11.1)	40.7(±11.3)	47.7(±10.3)
Normalized speed (m/s) †	-			
DRFT		0.3 (±0.1)	0.3 (±0.1)	0.4 (±0.1)#
DRFT with release		0.3 (±0.1)	0.3 (±0.1)	0.3 (±0.1)#
Normalized stride length (m) †	-			
DRFT		0.3 (±0.1)	0.3 (±0.1)	0.4 (±0.1)*#
DRFT with release		0.3 (±0.1)	0.3 (±0.1)	0.4 (±0.1)

Legend: DRFT: distal rectus femoris transfer, **E0**: preoperatively, **E1**: 1 year postoperatively, **E2**: 9 years postoperatively, **†**These parameters are normalized to body height. * Significant difference from E0. # Significant difference from E1.

4.3.2.2. Kinematics

The kinematics changes are presented in the table 25 and in the figure 23 by average sagittal plane kinematic mass graphs.

There was found an increase of the APT at E1 in both groups (Fig.24A). This increase was significant only in the DRFT group and there was a significant group difference at 1 year postsurgical. The APT decreased at E2 in both groups and nearly baseline status was reached. At long-term follow-up there was no significant group difference, indicating that function was comparable in both groups.

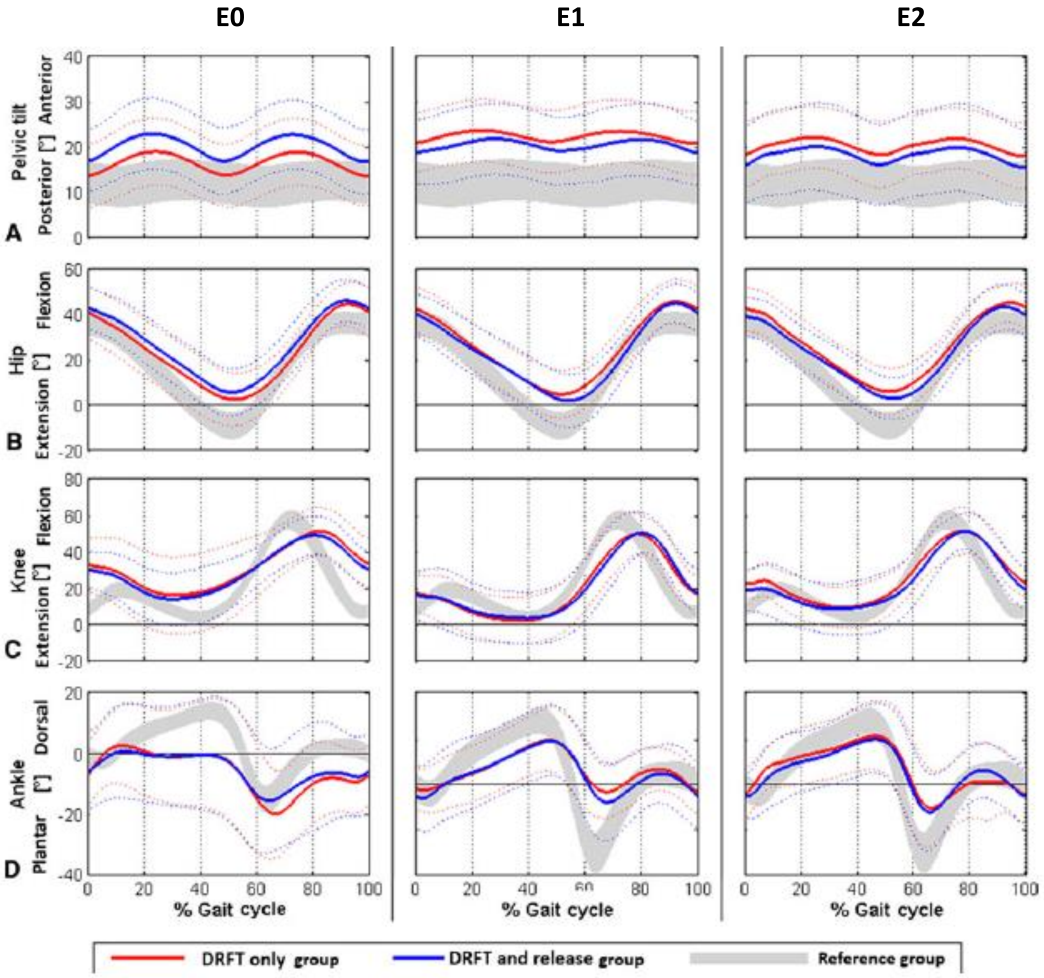
Knee ROM in swing increased significantly in both groups at E1 with no significant group differences (Fig.24B). The knee ROM in swing had decreased in both groups without any group differences between E1 and E2. However, this deterioration was only significant for DRFT with release group. Knee flexion velocity increased significantly in both groups with no significant group difference at E1. At E2 it was decreased in both groups, but it was only significant in DRFT group (no group difference). Peak knee flexion in swing improved by approximately 3° in both groups 1 year after surgery and at long-term follow-up in DRFT group there was found a slight decrease, whereas a slight increase in the DRFT with release group. However, these changes were neither statistically significant nor was there a significant group difference. The timing of peak knee flexion improved significantly in both groups 1 year postoperatively and remained without any group difference up to the long-term follow-up. The minimum knee flexion in stance decreased significantly in both groups at E1 (no group difference) and a recurrence was found at E2 for both groups, whereas this recurrence was significant only in DRFT with release group (no group difference).

25. Table: Outcomes of kinematics parameters (120)

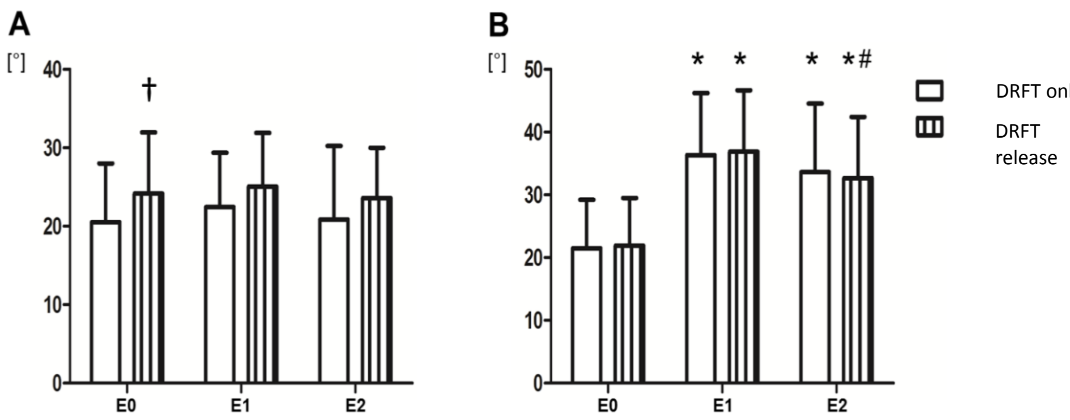
DOI:10.14753/SE.2016.1896

	Normal reference	E0	E1	E2	Group diff.
Max pelvic tilt (°)	13 (±5)				E0
DRFT		21 (±7)	22 (±7)	21 (±9)	
DRFT with release		24 (±8)	25 (±7)	24 (±6)	
Mean pelvic tilt (°)	12 (±5)				E0, E1
DRFT		16 (±7)	19 (±7)*	17 (±9)	
DRFT with release		20 (±7)	22 (±7)	20 (±7)	
Max hip flexion in stance (°)	34 (±6)				
		40 (±11)	39 (±8)	39 (±9)	
		43 (±9)	42 (±10)	42 (±10)	
Mean hip flexion in stance (°)	9 (±6)				E0
DRFT		16 (±10)	16 (±10)	17 (±8)	
DRFT with release		21 (±9)	19 (±9)	20 (±9)	
Peak knee flexion in swing (°)	56 (±6)				
DRFT		52 (±12)	55 (±8)	53 (±10)	
DRFT with release		51 (±10)	54 (±9)	55 (±8)	
Timing of peak knee flexion (%GC)	72 (±1)				
DRFT		81 (±4)	79 (±5)*	78 (±3)*	
DRFT with release		80 (±5)	78 (±5)*	78 (±5)*	
Knee flexion velocity (°/%GC)	1.6 (±0.2)				
DRFT		0.8 (±0.4)	1.2 (±0.4)*	1.0(± 0.5)*#	
DRFT with release		0.8 (±0.3)	1.2 (±0.4)*	1.1 (±0.4)*	
Knee ROM (°)	53 (±5)				
DRFT		40 (±15)	54 (±13)*	45 (±14)*#	
DRFT with release		38 (±13)	52 (±13)*	47 (±13)*#	
Knee ROM in swing (°)	50 (±5)				
DRFT		22 (±8)	37 (±10)*	34 (±11)*	
DRFT with release		22 (±8)	37 (±10)*	33 (±10)*#	
Min knee flexion in stance (°)	4 (±4)				
DRFT		13 (±19)	2 (±13)*	8 (±13)	
DRFT with release		13 (±14)	1 (±13)*	8 (±11)#	

Legend: DRFT: distal rectus femoris transfer, **E0**: preoperatively, **E1**: 1 year postoperatively, **E2**: 9 years postoperatively * Significant difference from E0. # Significant difference from E1.



23. Figure: Average sagittal plane kinematic mass graphs are shown for pelvic tilt, hip flexion, knee flexion, and ankle dorsiflexion and plantar flexion for all patients in the DRFT only group (red line), the DRFT and release group (blue line), and for an age-matched group of 48 subjects (gray area) (including 1 SD). The dotted red and blue lines correspond to ± 1 SD. Graphs are presented for all preoperative (E1), 1-year postoperative (E1), and 9- year postoperative (E2) examinations. Positive values indicate anterior pelvic tilt, hip flexion, knee flexion, and ankle dorsiflexion (120).



24. Figure: The graphs show the mean and SD for (A) maximum pelvic tilt before surgery (E0), 1 year after surgery (E1), and 9 years after surgery (E2), and (B) knee ROM in swing before surgery (E0), 1 year after surgery (E1), and 9 years after surgery (E2). * Significant difference from preoperative; #significant difference from 1 year postoperative; †differences between DRFT group versus DRFT and release group (120).

4.4. Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendinous lengthening in children with spastic diplegia

4.4.1. Clinical examinations

Thomas test and popliteal angle have improved significantly 1 year after surgery in both groups but it deteriorated significantly at E2 (Tabl.26). Passive knee extension has increased significantly at E1 in both groups. At E2 was found a slight decrease in both groups, but it was not significant. These 3 parameters have not showed any group differences by examinations. In both groups the passive ankle dorsiflexion improved significantly 1 year postsurgical but it was not maintained. At long-term follow-up the passive ankle dorsiflexion deteriorated significantly in both groups there was a group difference.

26. Table: Outcomes of clinical examinations (10)

	E0	E1	E2	Group difference
Thomas test (°)				
CBM	15 (±10)#§	5 (±7)*§	9 (±7)*#	
MTL	12 (±13) #§	5 (±6) *§	6 (±7) *#	
Popliteal angle (°)				
CBM	48 (±17)#	22 (±14) *§	54 (±16)#	
MTL	51 (±21)#	32 (±18) *§	53 (±15)#	
Passive knee extension (°)				
CBM	-7 (±6) #§	0 (±2)*	-2 (±8)*	
MTL	-9 (±13) #§	-1 (±6)*	-1 (±6)*	
Passive ankle dorsiflexion (°)				
CBM	-1 (±13)#	7 (±8) *§	5 (±7)#	E2
MTL	-0.4 (±14)#	4 (±7) *§	0.4 (±8)#	
Passive ankle plantar flexion (°)				
CBM	38 (±5)#	39 (±6) *§	34 (±10)*	E1, E2
MTL	43 (±11)#	35 (±11)*	35 (±18)#	

Legend: * Significant difference from E0. # Significant difference from E1. § Sinificant different from E2.

4.4.2. Global gait variables

The GMFCS levels have not changed significant over the years in either groups and there was no significant difference in the GMFCS changes between the 2 groups: CBM and MTL.

The GGI reduced one year after surgery in both groups. The improvements of GGI were maintained in the long-term follow up, but a significant increase showed in the CBM group, without any group difference (Tabl.11).

4.4.3. 3D gait analysis

4.4.3.1. Temporal parameters

Normalized cadence was significantly reduced at E1 in both groups, while this decrease was larger in CBM group (Tabl.27). However both groups showed a significant improvement at E2, the values were higher in MTL group. Normalized walking speed was significantly reduced in both groups 1 year postoperatively, but recovered and no differences were found between the two groups at E2. Concerning normalized step length, there was no significant difference between the examinations and groups.

27. Table: Outcomes of temporal parameters (10)

	E0	E1	E2	Group difference
Normalized cadence (steps/min)				E2
CBM	43.3 (±8.7)#	35.5 (±10.6)*§	41.8 (±9.7)#	
MTL	42.9 (±5.6)#	37.9 (±10.8) *§	45.6 (±6.2)#	
Normalized speed (m/s)				
CBM	0.22 (±0.07)#	0.19 (±0.08) *§	0.20 (±0.08)#	
MTL	0.22 (±0.06)#	0.19 (±0.09) *§	0.22 (±0.05)#	
Normalized step length (m)				
CBM	0.30 (±0.07)	0.30 (±0.07)	0.28 (±0.05)	
MTL	0.31 (±0.05)	0.29 (±0.08)	0.28 (±0.05)	

Legend: CBM: conversion of biarticular muscles to monoarticular muscles, **MTL:** muscle tendon lengthening surgery, **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 9 years postoperatively
* Significant difference from E0. # Significant difference from E1. § Significant different from E2.

4.4.3.2. Kinematics

The results are summarized in Table 28 and Fig. 25.

Pelvic tilt significantly increased ($p<0.01$) in MTL group directly after surgery while there was only a slight but not significant increase in the CBM group. At long-term follow-up both groups showed a decrease and ended up in identical pelvic tilt, there was no significant difference between the two groups at E2. However, compared to baseline, pelvic tilt in MTL group was still significantly increased (p=0.04). Minimum hip flexion in stance was only decreased in CBM group, but this decrease was not significant. At long-term follow-up both groups showed identical minimum hip flexion in stance.

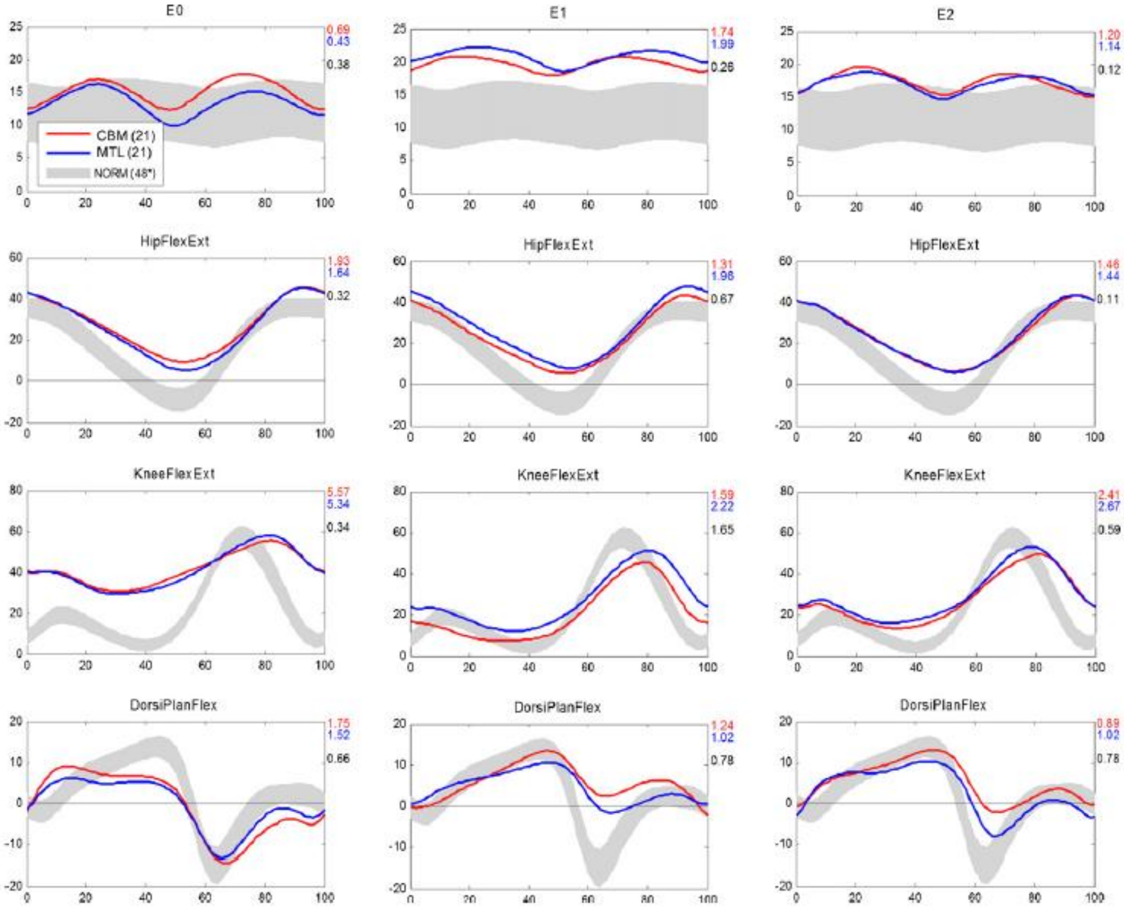
Range of knee flexion was increased significantly at E1 in both groups without significant group differences, but it was significantly deteriorated at long-term follow-up in both groups. Minimum knee flexion in stance was significantly decreased in both groups, while CBM group showed significantly more of a decrease. Knee flexion at initial contact was significantly decreased in both groups after surgery and a significant increase was found in both groups at long-term follow-up. In both groups peak knee flexion in swing showed a significant decrease at E1 (significant group difference), but this change was greater in CBM group. However at E2 a slight increase in peak knee flexion in swing in CBM group was found, there was still a group difference. In both groups significant deterioration of both stance phase parameters was found between E1 and E2, and the 2 groups showed similar values for all kinematic knee parameters at E2, with the exception of peak knee flexion in swing, where a difference was found.

Concerning ankle kinematics, range of dorsi/plantar flexion was significantly reduced in both groups at E1 and only partial-yet significant –recovery was found in both groups at E2. Peak ankle dorsiflexion in stance was improved significantly in both groups at E1 and it was maintained at E2 with significant group difference at the same examination.

28. Table: Outcomes of kinematics (120)

	E0	E1	E2	Group difference
Mean pelvic tilt (°)				
CBM	15 (±6)	19 (±7)§	17 (±8)#	
MTL	14 (±8)#§	21 (±8)*§	17 (±7)*#	
Minimum hip flexion in stance (°)				
CBM	10 (±15)	4 (±10)	6 (±11)	
MTL	6 (±11)	6 (±14)	7 (±11)	
ROM of knee flexion/extension (°)				
CBM	30 (±13)#§	44 (±11)*§	39 (±12)*#	
MTL	33 (±13) #§	48 (±12) *§	40 (±11) *#	
Knee flexion at initial contact (°)				E1
CBM	41 (±14) #§	16 (±10) *§	24 (±8) *#	
MTL	41 (±16) #§	23 (±6) *§	27 (±8) *#	
Minimum knee flexion in stance (°)				
CBM	28 (±20) #§	5 (±14) *§	13 (±11) *#	
MTL	28 (±21) #§	7 (±13) *§	16 (±10) *#	
Peak knee flexion in swing (°)				E1, E2
CBM	58 (±12) #§	49 (±10)*	52 (±9)*	
MTL	61 (±13) #§	56 (±8)*	56 (±7)*	
ROM of dorsi/plantar flexion (°)				
CBM	29 (±13) #§	20 (±7) *§	21 (±7) *#	
MTL	25 (±8) #§	18 (±5) *§	23 (±7) *#	
Peak dorsiflexion in stance (°)				E2
CBM	9 (±18) #§	14 (±8)*	15 (±6) *	
MTL	8 (±13) #§	12 (±6) *	12 (±7) *	

Legend: CBM: conversion of biarticular muscles to monoarticular muscles, **MTL:** muscle tendon lengthening surgery, **E0**: preoperative, **E1**: 1 year postoperatively, **E2**: 9 years postoperatively
* Significant difference from E0. # Significant difference from E1. § Significant different from E2.

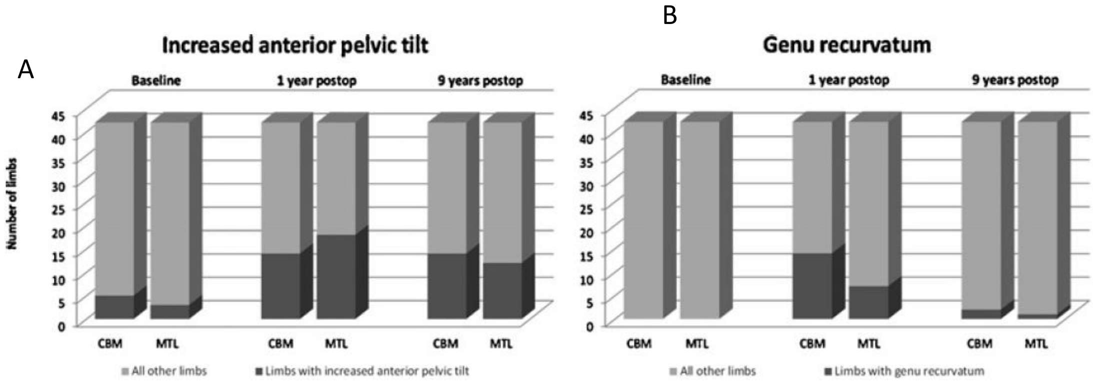


25. Figure: Sagittal plane kinematics (10). Average sagittal plane kinematic mass graphs of pelvic tilt (1st line), hip flexion (2nd line), knee flexion (3rd line) and ankle dorsi/ plantar flexion (4th line) for all patients of the CBM group (red line) and the MTL group (blue line) as well as for an age-matched group of 48 norm subjects, represented by the grey area (including 1 standard deviation), are shown. Graphs are visualized for all examinations: preoperative (E0, 1st column), 1 year post- (E1, 2nd column) and 9 years postoperative (E2, 3rd column). Positive values indicate anterior pelvic tilt, hip flexion, knee flexion and ankle dorsiflexion. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

4.4.4. Adverse effects and recurrence of flexed knee gait

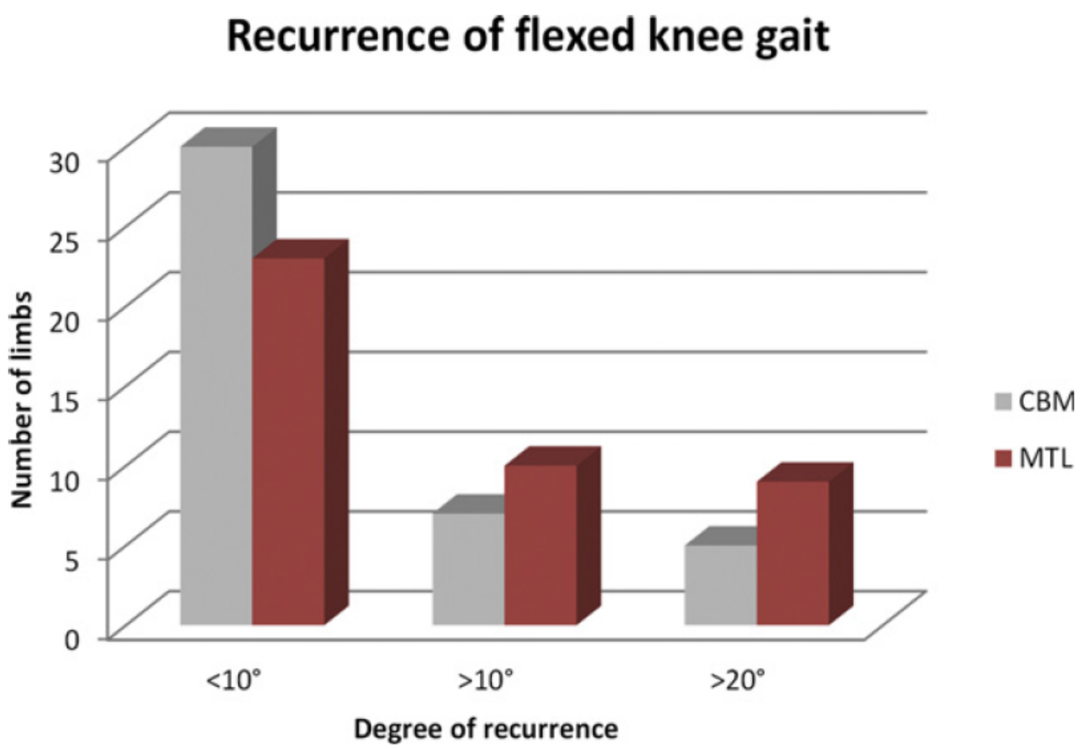
An APT value more than 2 SD above the norm was classified “increased”. One year after surgery 33% of the limbs in the CBM group and 43% of the limbs in the MTL group showed increased APT at E1. This increase of pelvic tilt decreased between E1 and E2 in the MTL group, while it did not change in the CBM group, resulting in almost identical prevalence of increased pelvic tilt in 2 groups at E2 (Fig.26A).

At E1 33% of the limbs (14 out of 42: 5 bilateral, 4 unilateral) in the CBM group and 17% (7 out of 42: 2 bilateral, 3 unilateral) of the limbs in the MTL group showed genu recurvatum (knee hyperextension of >5° in stance phase). At long-term follow-up in both groups genu recurvatum was found only in 5% of the limbs (2 unilateral) in the CBM group and only 2.5% (1 limb) in the MTL group (Fig.26B).



26. Figure: Adverse effects (10). The prevalence of increased anterior pelvic tilt (APT) and genu recurvatum among all limbs is shown for the CBM and the MTL group at the 1 year (E1) and the 9 year (E2) examination. Dark grey bars represent the affected limbs while light grey bars represent all other limbs. **CBM:** conversion of biarticular muscles to monoarticular muscles, **MTL:** muscle tendon lengthening surgery

The increase of minimum knee flexion in stance between E1 and E2 was the defination of recurrence of flexed knee gait. The calculation of them occurred for each limb in both groups. The next Fig.27 shows the distribution of it.



27. Figure: Recurrence of flexed knee gait (10). The amount of recurrence of flexed knee gait was calculated by the difference between minimum knee flexion in stance at E1 and E2 for each individual leg for all patients of both groups. According to this difference, the legs were categorized into three groups (abscissa): recurrence less than 10º (1st bar), recurrence of more than 10º (2nd bar), and recurrence of more than 20 º (3rd bar). The number of legs is shown on the axis of ordinates.

5. Discussion

5.1. Development of knee function after hamstring lengthening as a part of multilevel surgery in children with spastic diplegia

Various inconsistent short-term outcome studies have demonstrated improvement in terms of knee extension during stance phase and the popliteal angle following hamstring lengthening (26, 38, 41, 87-92). Comparable initial results in terms of the findings of gait analysis and the popliteal angle were found following intramuscular hamstring lengthening in the present study, corroborating these previous findings.

Because of CNS damage and growth, subsequent changes in gait pattern and joint function are expected after multilevel surgery. Some investigations demonstrating results after more than three years of follow-up are rare. The findings of Adolfsen et al. showed a maintained improvement at four years after intramuscular hamstring lengthening in six patients with CP (27). Chang et al found an increase in the popliteal angle after four years but no deterioration in kinematics (87). Gordon et al. reported maintained improvement in the popliteal angle and peak knee extension in stance at a mean of 2.8 years after medial hamstring tenotomy in a study of twenty-nine patients with CP (91). Rodda et al. reported maintained improvement in the popliteal angle and minimum knee flexion in stance in a five-year outcome study of ten patients with CP (31). The authors reported a slight but not significant increase in minimum knee flexion in stance.

In the present study, outcome was evaluated an average of eight years (range, 6 to 12 years) postoperatively, and most patients were skeletally mature at the time of long-term follow-up because the average age at the time of surgery was ten years. The period between the ages of ten and eighteen years, in which further changes in gait may develop because of growth, is crucial for long-term outcome. In contrast to all of the intermediate-term studies discussed above, a significant deterioration of dynamic parameters, including mean knee flexion in stance, knee flexion at initial contact, and minimum knee flexion in stance, was found at long-term follow-up in the present study. Surprisingly, the improvements in the GGI and in the GMFCS level were maintained, indicating that overall functional results after SEMLS are preserved over the long-term. However, both indices are global variables, which do not take into account detailed function of specific joints. Because the knee joint plays a central role in the gait of patients with CP, the deterioration in knee kinematic parameters should be considered when planning the treatment of flexed knee gait. Additional lateral hamstring lengthening was needed in nine patients (18 limbs) to achieve adequate correction intraoperatively. Subgroup analysis showed that the medial and combined medial and lateral hamstring lengthening groups had identical sagittal knee parameters at long-term follow-up, but at the cost of increased pelvic tilt in the combined medial and lateral hamstring lengthening group.

The deterioration of the popliteal angle was noted in the present study supports the findings of previous studies (121). Thompson et al. proposed that the aim of flexed knee gait correction is primarily to improve function and not simply to improve static parameters (114). The results of present study underline that the deterioration of the popliteal angle and the recurrence of flexed knee gait as shown on three-dimensional gait analysis do not correlate well. As previously shown for other components of gait in patients with CP, static clinical parameters are not useful as predictors of gait function (122-124). The popliteal angle is not suitable as a method for predicting outcome or as a preoperative indication for hamstring lengthening.

Various explanations for the recurrence of flexed knee gait should be considered as flexed knee gait represents a multiplanar problem and cannot be reduced to the knee joint only. Because of deterioration of the popliteal angle, recurrent hamstring tightness during growth represents one of the most frequently discussed possible causes. Furthermore, two different types of insufficiency may lead to recurrence over the years: proximal insufficiency (hamstrings) and distal insufficiency (lever arm dysfunction). Previous investigations have demonstrated the importance of muscle length to determine the dosage of lengthening and to avoid overcorrection (125-128). Hamstring lengthening may lead to unwanted functional effects such as increased pelvic tilt (active insufficiency of the hamstrings). This may explain the first increase in pelvic tilt, seen one year postoperatively in the present study. Increased pelvic tilt may be compensated by increased lumbar lordosis or by walking with the knees in a flexed position (compensatory flexed knee gait), which may contribute to the recurrence. Furhter shortening of the hamstrings raises the pelvis, which may explain the improvements in pelvic position between E1 and E2. However, flexed knee gait will increase pelvic tilt again. Decreased foot lever arm, which is caused by instability of the foot or increased external or internal tibial torsion, also may cause increased knee flexion. Instability of the foot (midfoot break) frequently is seen in pes valgus, which commonly accompanies flexed knee gait (70). Despite frequent osseus foot stabilization for the correction of pes valgus deformity in 53% of the limbs in the present study, flexed knee gait recurred.

Other reasons for a shortened foot lever arm are torsional deformities of the tibia, that were not treated during multilevel surgery (75, 78). To our knowledge, no clinical studies have evaluated the importance of increased external tibial torsion or midfoot break with regard to the recurrence of flexed knee gait. In the present investigation, tibial malalignment was treated with supramalleolar deterioration osteotomy in five limbs at the time of multilevel surgery.

Weakness of the quadriceps muscle is another possible factor that may cause or aggravate flexed knee gait. In the present study, distal rectus femoris transfer was performed on seventy-one limbs (91%). The rectus femoris muscle contributes about 12% of the total quadriceps muscle power and mass, which potentially is lost when distal rectus femoris transfer is performed (81). Furthermore, many patients with CP show continuous rectus femoris activity through the stance phase, which may lead to further weakness of the remaining vastii after distal rectus femoris transfer (83). Last, weight gain and increase in body mass index also may influence flexed knee gait and lead to recurrence (129).

Genu recurvatum is a common adverse effect of hamstring lengthening (27, 38, 87, 88). Adolfsen et al. identified genu recurvatum as a functionally relevant complication (27). In the report by Kay et al., the prevalence of genu recurvatum was higher in the group that was managed with combined medial and lateral hamstring lengthening (16%) (88). In the present study, genu recurvatum was found in 35% of the limbs, but there was no difference between the medial hamstring lengthening and combined medial and lateral hamstring lengthening groups. Knee hyperextension mainly occurs in patients with a preoperative jump knee gait pattern despite meticulous and successful equinus correction (Fig.20 , ankle kinematics), resulting in reduction of the excessive plantar flexion/knee extension couple. Therefore, this tendency for genu recurvatum seems to be attributed mainly to hamstring lengthening. Interestingly, the number of limbs with knee hyperextension decreased significantly, indicating that the knee hyperextension may resolve during the years of follow-up. However, 12% of the limbs showed residual genu recurvatum at the time of the long-term examination, and all were in patients who had a jump knee gait pattern preoperatively. Therefore, overcorrection represents a major complication, and a jump knee gait pattern should be taken into consideration before surgery. Another explanation for genu recurvatum at the time of long-tern follow-up may be a recurrence of equinus deformity. The treatment of genu recurvatum is a challenge, and strategies are limited. In the present investigation, patients who had genu recurvatum postoperatively used an ankle-foot orthosis to prevent knee hyperextension when walking long distances.

The proximal parts of the hamstring muscles are essential for hip extension and for stabilizing the pelvis (130). The increased pelvic tilt in the present investigation supports the results of previous studies and may represent one factor leading to recurrence (38, 87, 126). Alternative treatment strategies include the transfer of hamstring tendons to the femur to preserve the effects on the hip and pelvis (34, 106, 107). Unfortunately, only a handful of investigations have evaluated the short-term results after hamstring transfer, and, to our knowledge, no long-term reports exist (34, 106, 107). Another approach for correcting flexed knee gait is supracondylar femoral extension osteotomy used in combination with patellar tendon shortening in different investigations (131-134). Encouraging short-term results have been reported (33, 135). Thus, the combination of femoral extension osteotomy and patellar tendon shortening represents our current treatment strategy. However, this approach cannot be recommended without reservation because long-term studies are lacking.

There was a potential for selection bias in the present study that may limit the ability to generalize these results. In some cases children with CP may not be seen for follow-up after the age of sixteen years. This is one possible reason for the general lack of long-term reports. In the present study, sixty-eight patients were eligible for re-evaluation, of whom only thirty-nine (57%) returned for long-term follow-up.

Furthermore, the reported outcome may be influenced by other procedures that were performed at the time of multilevel surgery (24). Therefore, the results of the present study should be interpreted as a cumulative effect of multilevel surgery, with hamstring lengthening representing one central procedures for knee function.

5.2. Long-term results after distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff knee gait in spastic diplegic cerebral palsy

The treatment of stiff knee gait in CP is challenging and controversial because variable effects following distal rectus femoris transfer (DRFT) have been reported (39, 43, 99, 136-139). Since natural progression in patients with CP may lead to recurrence or aggravation of gait disturbances during growth, it is insufficient to consider only short-term outcome (40, 140). Only a few studies have previously investigated longer-term results following DRFT. Saw et al. reported maintained improvements in swing phase flexion but a significant loss of knee motion due to progressive crouch gait 4.6 years after DRFT in eighteen patients with CP (93). Limitations of that study were the small number of patients and the inclusion of different types of CP. In a comparative study, Moreau et al. showed benefits after DRFT in comparison with another cohort in which DRFT was not carried out (94). The positive effects of DRFT were maintained in a second evaluation 3 years after surgery.

Our study is the first, to our knowledge, to separately evaluate patients with normal or increased peak knee flexion in swing (commonly those with severe flexed knee contractures) who underwent DRFT as a prophylactic procedure to prevent peak knee flexion in swing (the P-DRFT group) and patients with decreased peak knee flexion in swing, for whom the aim was correction of decreased peak knee flexion in swing (the C-DRFT group). Mixing these two groups in previous reports may have masked improvements of peak knee flexion in swing after DRFT (39, 43, 99). In the present study, significant improvements in knee flexion in swing and knee flexion velocity one year after surgery were found for both groups, corroborating the results of previous investigations. However, peak knee flexion in swing and its timing were significantly improved in the C-DRFT group only, while peak knee flexion in swing was significantly decreased in the P-DRFT group and the timing did not change. The benefits in knee flexion in swing in the P-DRFT group seem to mostly occur because of the sizeable improvements of knee extension in stance phase. Despite prophylactic DRFT in the P-DRFT group, there was a mean decrease of 15° in peak knee flexion in swing, which means that the entire curve was shifted to downward. Prophylactic DRFT was not able to maintain peak knee flexion in swing within one standard deviation of the norm, but a comparable average value was noted in comparison with the C-DRFT group. However the question arises as to whether these effects can be attributed to the DRFT or whether SEMLS without prophylactic DRFT would lead to same results. In the patients with decreased peak knee flexion in swing preoperatively, the benefits in knee flexion in swing are mainly based on the mean improvements in peak knee flexion in swing (6°), which adds to the improved knee extension in stance phase, whereas the improvements in knee flexion in swing in the P-DRFT group are primarily the result of improved knee extension in stance. The increase of knee motion during swing phase in combination with an increased peak knee flexion in swing represents a benefit for foot clearance in swing phase and is great relevance for a stable, undisturbed, and smooth gait as approximately 60° of knee flexion is necessary for regular swing foot clearance (72). In the study by Hemo et al., conventional DRFT to the medial hamstrings was compared with DRFT attached to the iliotibial band, and no differences in the outcome were found between the techniques (95). The improvements found after DRFT are mainly related to the elimination of the rectus femoris as a knee extensor and the prevention of reattachment of the distal rectus tendon in its anatomic anterior position by transferring it to another site.

Our long-term follow-up showed that the improvements in knee kinematics could be maintained and even enhanced nine years after surgery in the C-DRFT group. The overall good results nine years postoperatively indicate that DRFT carried out as a part of SMELS is an effective approach for the treatment of stiff knee gait. However, contrary to the benefits in the C-DRFT group, the knee flexion in swing in the P-DRFT group deteriorated. This may be partially explained by the recurrence of flexed knee gait, which was significantly higher in the P-DRFT group. Since the correction of stance phase extension is the primary goal in these patients, the approach of performing DRFT as a prophylactic procedure is questioned because it weakens the knee extensors. This weakness may be one possible factor leading to recurrence of flexed knee gait in the P-DRFT group.

We showed that the patients with more involvement have more potential benefit from DRFT. A benefit for peak knee flexion in swing cannot be expected when the preoperative peak knee flexion in swing is >53°. Some patients showed a poor response to DRFT, and some developed recurrence of stiff knee gait. Different possible explanations should be considered. First, a persistent extensor moment may result from the transferred rectus muscle because of scar tissue or remaining extensor moment arm despite the transfer. This would underline the findings of Asakawa et al. and Riewald and Delp, who noted that the rectus femoris does not turn into a knee flexor and may have a persistent knee extensor moment (96-98, 141). This would also explain the persistence of the Duncan-Ely sign in our study. Second, increased muscle tone of the remaining quadriceps muscle may lead to a poor outcome in some patients. Various studies have tried to outline a precise outcome prediction for the DRFT (142-144). In a recent study, Reinbolt et al. developed a model for outcome prediction in consideration of different gait analysis variables and found a prediction accuracy of up to 80% (145). However, some uncertainty remains, potentially resulting in an unnecessary surgical procedure.

The question arises as to whether alternative treatment options such as distal release of the rectus femoris tendon are more effective. Sutherland et al. and Ounpuu et al. showed that rectus femoris release did not lead to a comparable or even better outcome in comparison with DRFT (44, 81). In a recent study, Cruz et al. reported a comparable outcome after intramuscular distal rectus recession and postulated that this procedure represents an alternative treatment strategy for the correction of stiff knee gait (146). However, peak knee flexion in swing and knee flexion in swing were not significantly improved. Recurrence after intramuscular lengthening is often seen in CP, and recurrence of stiff knee gait has to be expected (9). The long-term effects of this technique are unclear.

There are some limitations of the present investigation. A potential for selection bias in this study may have influenced the results. There were nine patients who could not be evaluated because they have moved or they declined participation. A potentially bad outcome in these patients may have had an influence on the results. However, this is a well-known problem of long-term studies. This study is the first, to our knowledge, to describe the long-term outcome at a mean of nine-years after DRFT in patients who were nearly all skeletally mature adults. Another important factor that may influence the development of clinical and kinematic parameters during the years is growth and increase of the body mass index. These influences on the outcome after multilevel surgery in general are not adequately addressed. Furthermore, concomitant procedures carried out during SEMLS may have an influence on knee kinematics. Some patients received an additional proximal rectus femoris release for the correction of hip flexion contracture and double-bump pattern of the pelvis on gait analysis. The possible influence of proximal rectus release on knee kinematics, which has been reported to be limited, should be considered when interpreting the outcome in this study (100).

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5.3. Does proximal rectus femoris release influence kinematics in patients with cerebral palsy and stiff knee gait?

An injury of the high-level motor control in patients with CP affects the mechanisms of biarticular muscles considerably (147-150). The rectus femoris muscle, which was found to act independently of the vasti, is one of the biarticular muscles that is commonly involved in patients with CP and causes stiff knee gait by reducing knee flexion during that swing phase of gait (151). It is believed to cause or aggravate increased APT. DRFT is commonly performed to correct stiff knee gait, however variable outcomes have been reported (9, 43, 93, 94). A proximal rectus femoris release was done to eliminate rectus function at the pelvic level and to influence knee motion but was found to be less effective than DRFT. However, to our knowledge, the outcome after a combination of both procedures has not been reported. In our long-term study we could not find any beneficial effect of proximal rectus femoris release on knee and pelvic kinematics and the indication for this procedure in addition to DRFT should be questioned.

Our study had numerous limitations. First, this was a retrospective study, which may influence the comparability of the two groups chosen, especially concerning the preoperative indication for selected surgical procedures. Furthermore, the patients in our two study groups had several additional procedures (8 procedures per subject on average; Tabl.10) as a part of SEMLS. Some concomitant procedures affect knee kinematics, such as hamstring lengthening, which improves knee motion in the long-term (9, 152). Hamstring lengthening may increase APT, since the hamstrings stabilize the pelvis and lengthening may lead to insufficiency (9). However, hamstring lengthening was done in a nearly equal number of patients in both groups. There were more patients who received a proximal femoral osteotomy in the group with additional rectus femoris release compared with the group with DRFT only. This may have influenced the results. Because proximal rectus femoris release also was done to reduce increased APT, the group with additional rectus femoris release had a significantly higher (p=0.03) mean pelvic tilt at baseline compared with the DRFT only group. This may bias a comparison of pelvic tilt development between the groups. In this study only sagittal plane kinematics were evaluated. Potential changes in frontal and transversal plane kinematics are not reported. Finally, gait analysis systems are subject to measurement errors owing to inherent system characteristics and marker placement variability. Ounpuu et al and Sutherland et al.

reported that proximal rectus femoris release reduces hip flexion contracture and lumbar lordosis but also improves knee flexion in swing (43, 82). Since it was shown that DRFT leads to a superior outcome compared with proximal release, it has become the gold standard. There are numerous reports regarding the treatment of stiff knee gait using DRFT or proximal rectus femoris release (9, 42-45, 81-83, 93-95, 119, 145, 152, 153). Good initial results were reported (42-45, 81-83, 95, 145, 152, 154). However, some authors have reported discrepant outcomes after DRFT, and a poor or no response rate of approximately 20% was found in a recent long-term investigation (93, 94, 119, 153). These inconsistent outcomes may be explained by a persistent extensor moment of the rectus femoris after transfer (96, 97, 155). In such cases an additional proximal rectus femoris release may have an additional beneficial effect on knee motion.

According to our findings, the kinematic knee parameters (range of flexion in swing phase, peak knee flexion in swing, and knee flexion velocity) were significantly improved in both groups independent of whether rectus release was performed. The short-term improvements were comparable to those reported in the literature (43, 44, 81, 94, 153). The effects in the DRFT only group of our study were sustained at the long-term follow-up, with the expectation of knee ROM in swing corroborating the findings of previous studies investigating long-term results after rectus femoris transfer (93, 94, 119, 153). Knee ROM in swing decreased significantly in both groups but mainly as an indirect effect of recurrence in the stance phase knee flexion. When comparing the outcome of the DRFT only group with the group that received the additional proximal rectus femoris release, we could not find any group differences 9 years after surgery. Therefore, the results of our study indicate that the short- and long-term influences of proximal rectus femoris release on DRFT effects on the knee are negligible.

Two possible interpretations of our findings should be considered: (I) if DRFT effectively removes the knee-extending function of the rectus femoris, no surgery on the proximal part should directly influence knee kinematics. In this case our results would underline the effectiveness of DRFT in correcting stiff knee gait by eliminating the rectus femoris muscle (42-45, 81, 83, 93-95, 119, 145, 152, 153). However, since the rectus femoris may secondarily influence knee motion by generating an internal hip flexion moment and a secondary external knee flexion moment, the proximal rectus femoris release is potentially able to influence the effects of DRFT on the knee (101). Our results that there were no differences between those patient who received additional proximal release and those who did not may indicate that the proximal rectus femoris release does not relevantly influence this mechanism. (II) In cases of persistent internal knee extensor moment of the rectus femoris on the knee after DRFT, additional surgery in the proximal part does not affect knee function (96, 97, 155). Therefore, although clearly combined in biomechanics, clinically hip flexion and knee extension appear to be independent functions of the rectus femoris.

Causes of increased pelvic tilt can be a shortened rectus femoris or iliopsoas or/and weak hip extensors and poor selective motor control of the gluteus muscles and high femoral anteversion. Proximal rectus femoris release is performed to improve hip extension and to reduce increased APT. There were significant group differences in the maximum and mean pelvic tilt before surgery, reflecting the indication for an additional proximal rectus femoris release in this group. This group difference remained at the 1-year follow-up for mean pelvic tilt, but at long-term evaluation no significant difference between the two groups was found. The pelvic tilt increased slightly in both groups after surgery and this increase should be seen mainly as the result of concomitant hamstring lengthening on many patients in both groups (9). Therefore, the proximal rectus release did not prevent APT from increasing, corroborating the findings of McMulkin et al. (100). This is underlined by the hip kinematics with no significant changes in hip motion being observed in either group. This is in accordance with the findings of Sutherland et al. (81). We believe that surgery on the proximal parts of rectus femoris does not relevantly influence pelvic and hip kinematics. It is a relevant question why we performed only a few psoas lengthening procedures (156). We attempted to avoid psoas lengthening so as not to cause propulsion problems. It was done only if we were unable to correct a hip flexion contracture intraoperatively during proximal rectus femoris release. It seems that increased hip flexion resolves after the correction of distal problems and this is in accordance with the results of Rutz et al. (157). However, this needs to be investigated further.

5.4. Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendinous lengthening in children with spastic diplegia

Since a high level of motor control is needed for normal function of the biarticular muscles of the lower extremity, patients with CP tend to have more difficulty controlling biarticular than monoarticular muscles (34, 103, 148). The disturbed function of the biarticular muscles in CP is of major importance in development of gait abnormalities such as flexed knee gait. These abnormalities in the function of biarticular muscles can be modified by MTL as a component of SEMLS, and various studies have reported satisfactory results after hamstring lengthening (9, 26, 38, 57, 87, 88, 91). However, increased APT due to hamstring weakness and recurrence of flexed knee gait have been as adverse effects (9, 38, 88, 91). This may partially be explained by the fact that some patients with flexed knee gait have normal or even increased hamstring length (125). Hamstring lengthening in these patients may lead muscle weakness and APT. Increased APT has been suggested to be a major factor to leading to recurrence of flexed knee gait (9). CBM aims to reduce these adverse effects, but its surgical impact is considerably higher than that of MTL.

In our study CBM resulted in a better overall kinematic outcome than MTL 1 year after surgery, reflected in less APT, greater improvement of knee extension in stance and a smaller popliteal angle. Despite concomitant lengthening of semimembranosus and in a few cases biceps femoris there was no significant increase in APT in the CBM group, while the patients of the MTL group showed a significant increase in mean APT. It seems that the transfer of the semitendinosus tendon has the potential to stabilize the pelvis. The short-term efficiency of CBM is in accordance with previous reports (34, 107). However, most of patients in past studies were not followed up after their main growth spurt and changes beyond the scope of these studies may influence the outcome, since deterioration of gait parameters during growth is possible in children with CP (140). It is therefore of major interest whether these advantages of CBM are maintained until the end of growth.

These favourable effects of CBM in our study vanished at long-term follow-up, resulting in comparable overall kinematic and clinical function in the two groups. This shows that CBM, a significantly more extensive intervention for the patient and surgeon, has no long-term advantage over MTL. When considering each individual patient, the prevalence of increased APT was higher in the MTL group than in the CBM group 1 year after surgery. This may be explained by the weakening and over lengthening of the hamstring in the MTL group, while it seems that the transfer of the semitendinosus tendon to the femur has some stabilizing effect on the pelvis in the CBM group. However, the prevalence of increased APT in both groups is excessively high and cannot be left unremarked. Both procedures are accompanied by significant weakening of the hamstrings, leading to loss of pelvic stability in the sagittal plane. This increase in APT and the conversion of the gastrocnemius muscle may explain why the CBM group tended to display genu recurvatum 1 year after surgery more often than the MTL group. Genu recurvatum was significantly less frequent at long-term follow-up, when both groups showed very low prevalence of knee hyperextension. However, the prevalence of increased APT did not change between E1 and E2 in the CBM group and only a marginal reduction was found in MTL group. These long-term results indicate that a substantial increase in mean APT has to be expected after any surgery, only partially recovering over time. Since aponeurotic semimembranosus lengthening was done in all patients of both groups and lateral hamstrings lengthening was needed in a few cases, persistent hamstring over length and weakness has to be taken into consideration as the major cause for increased APT. Persistent increased APT should be considered as one possible factor leading to partial recurrence of flexed knee gait as found in this study (9). However, since the overall APT improved in the MTL group, some patients must have the potential to recover muscle strength and sufficiently stabilize the pelvis despite treatment by MTL, or muscle length is re-established as an effect of growth. It may therefore be meaningful to ask which patients are prone to develop increased APT. In accordance with the suggestion of Ma et al. there is a general belief that patients with higher GMFCS level, who are limited concerning compensation of muscle weakness, are particularly prone to adverse effects and recurrence of flexed knee gait (107). The patients in the present study were also matched according to GMFCS level in order to compare patients with the same degree severity. We found no correlation between GMFCS level and development of increased APT. However, this may be due to the relatively small sample size. With regard to recurrence of flexed knee gait, both groups showed a significant deterioration between the 1-year and long-term examinations, while the MTL group exhibited a non-significant tendency towards greater deterioration. The long-term recurrence of flexed knee gait seems to be influenced by increased APT. Since CBM did not yield superior results and its surgical impact in the patients is considerably higher than that of MTL, CBM cannot be recommended.

One recently described treatment concept for flexed knee gait correction comprises femoral extension osteotomy and patella tendon shortening (135). In the absence of long-term results, however, this procedure cannot yet be compared with MTL and CBM.

During SEMLS different surgical procedures are commonly performed in one session. Therefore, the concomitant procedures may have affected the results in both patient groups in our study. Although we strove to minimize these influences by matching the patients according to different gait analysis parameters, this factor should be considered when interpreting the results.

6. Conclusion

6.1. Development of knee function after hamstring lengthening as a part of multilevel surgery in children with spastic diplegia

For the treatment of flexed knee gait, intramuscular hamstring lengthening as a part of multilevel surgery provides satisfactory short-term improvements in knee function during gait. However, the results of the present long-term study showed a significant recurrence of flexed knee gait. Increased pelvic tilt, found in approximately 50% of the limbs postoperatively, may be one factor leading to recurrence. Genu recurvatum, a possible consequence of overcorrection with hamstring lengthening, was seen frequently at one year postoperatively. Genu recurvatum may disappear over the years, but it remains a functional problem for patients with a preoperative jump knee gait pattern. Careful selection for surgery and appropriate hamstring lengthening are needed for those patients. Knowledge about the long-term effects and possible recurrence is of greatest interest when planning treatment and should be considered when providing preoperative information to patients and their parents. Extension osteotomy of the femur in combination with patellar advancement as a current alternative treatment strategy has led to encouraging short-term results, but it remains to be seen if these short-term results are maintained at long-term follow-up (33, 135).

6.2. Long-term results after distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff knee gait in spastic diplegic cerebral palsy

In conclusion, DRFT leads to long-lasting improvements of peak knee flexion in swing, timing of peak knee flexion in swing and range of knee flexion in swing in patients with preoperatively decreased peak knee flexion in swing, although patients with more involvement had the most improvement. DRFT as a part of SEMLS has the potential to counteract the natural progression of knee function limitation in patients with spastic diplegia. However, since 18% of the patients showed a permanently poor response, DRFT has no influence in some patients, and different mechanisms may be responsible for recurrence, which was found in 15% of the patients. When patients with severe knee flexion contractures had prophylactic DRFT, a significant loss in peak knee flexion in swing was noted. SEMLS without DRFT may have resulted in the same outcome, and it is questionable whether DRFT, which weakens knee extensor power, is indicated in these patients, for whom improvement of knee extension is the primary aim of treatment.

6.3. Does proximal rectus femoris release influence kinematics in patients with cerebral palsy and stiff knee gait?

We evaluated the effects on the knee and pelvis when DRFT and proximal rectus femoris release are performed simultaneously. Our short- and long-term results suggest that a combination of the two procedures does not further improve the effects of DRFT on stiff knee gait. Furthermore, we found no superior outcome concerning APT. Any additional effects of a concomitant proximal rectus femoris release on the knee and pelvis therefore are negligible.

6.4. Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendinous lengthening in children with spastic diplegia

CBM as an alternative treatment option for the correction of flexed knee gait. CBM and MTL lead to increase in mean APT 1 year after surgery, but it was significant only for MTL group. The lower prevalence of increased APT in CBM group may be based on positive effect of semitendinosus on pelvic stability. CBM shows a better outcome one year after surgery on knee kinematics in comparison to conventional MTL. 9 years after surgery an identical average outcome is found in both groups. These results demonstrate that more extensive procedure of CBM has no long-term benefit over MTL.

7. Summary

The most common gait disorders in patients with cerebral palsy are flexed knee gait and stiff knee gait, commonly treated by single event multilevel surgery (SEMLS). SEMLS is aimed at the correction of all deformity in every plane and in one level surgery. Different bony and soft tissues will be operated. For indication and control after SEMLS the 3D gait analysis as objective and dynamic technique became accepted. Our investigations have presented a long-term follow-up (mean 9 years) when most patients were skeletally mature. Intramuscular hamstring lengthening to correct flexed knee gait maintained good short-term results, but in long-term follow-up showed a deterioration of knee extension. An increased anterior pelvic tilt is a common complication of hamstring lengthening and it persisted in the long-term follow-up. The hamstring lengthening to correct flexed knee gait is a not suggested technique. According to our comparative study, the transfer of semitendinosus muscle tendon to femur does not result in a better outcome in long-term follow-up. An alternative technique should be distal extension osteotomy with patellar tendon advancement, but the long-term results are not yet available. Patients with jump-knee gait had to be discouraged from hamstring lengthening, because of the high prevalence of lasting genu recurvatum. To correct stiff knee gait by SEMLS with additional DRFT showed significantly better flexion in swing phase after operation. Patients with preoperatively diminished maximal knee flexion in swing benefited from DRFT. However patients with flexed knee gait had degraded the knee extension after DRFT. The prophylactic DRFT on flexed knee gait patients had to be discouraged. It could be performed secondarily, if it is necessary, by metal displacement. In long-term outcome the positive effects of DRFT by stiff knee gait must be evaluated. The results of the study about the influence of the proximal rectus femoris release on the knee kinematics on patients with stiff knee gait pattern show that by applying proximal rectus femoris release, the effect on the kinematics and pelvic tilt of DRFT in children with CP are negligible.

SEMLS showed an effective procedure to correct the gait disorders on children with cerebral palsy. Utilizing long-term follow-up, permanent effects, deteriorations, over- and under corrections could be established. These objective parameters are of great value in planning the correct procedure and to communicate with patients and their parents.

8. Összefoglalás

A cerebral paretikus gyerekek leggyakoribb járási deformitásai közé tartozik az úgy nevezett guggoló (flexed knee gait), illetve a merev térddel (stiff knee gait) való járás. Ezeket legtöbbször többszintű műtéttel (SEMLS) kezelik. Ez annyit jelent, hogy egy operáció során akár több ízület is érintve lehet, illetve mind lágy- és csontszövet is operálásra kerülhet. A műtéti indikáció, illetve a műtétel sikerének kontrolljára a 3D járásanalízis mára már elfogadott módszer. A kutatásaink hosszú távú utánkövetéseket mutatnak be, amikor a betegek nagy részének csontjainak érési folyamata lezárult.

A hajlított térddel jár betegek járásának intramusculraris hamstringhosszabítása jó rövidtávú eredményeket mutatott, azonban a hosszú távú utánkövetés a térd extensiójának romlását jelezte. A fokozott medence előrebillenés (increased anterior pelvic tilt) általános következménye a hamstringek hosszabbításának, mely hosszú távú vizsgálatoknál is látható volt. Az eredményeinknek köszönhetően megállapíthatjuk, hogy a hamstringek hosszabbítása a hajlított térddel való járás esetében nem javasolt. A másik összehasonlító tanulmányunk, melyben a semitendinosus izomnak inának áthelyezése történt a femurra, sem adott jobb eredményeket hosszú távon. Ezzel szemben azonban egy alternatív technika lehet a distalis extensios osteotomia a lig. patellae áthelyezésével egybekötve, azonban ennek hosszú távú eredményei mindezidáig nem kerültek publikálásra. Emellett fontos kimutatásunk, mely alátámasztja mások eredményeit is, hogy azon betegeknél, akik úgynevezett ugró térddel (jump knee gait) járnak, semmiképpen sem szabad elvégezni a hamstringek hosszabbítását, hiszen ez nagy eséllyel hosszú távon is fennmaradó, mondhatni kezelhetetlen genu recurvatumot eredményez.

A merev térddel járók esetében azon csoportnál, melynél a SEMLS során DRFT (distalis rectus transzfer) is történt, szignifikánsan jobb térdhajlítás volt látható a lengési fázis során. Két csoportra bontva őket: azon betegeknek, melyeknek preoperatív csökkent volt a maximalis térdflexiójuk a lengési fázisban, előnyük származott a DRFT-től. Azonban azon betegeknek, akik guggoló térddel jártak, s csupán profilaktikus beavaztkozásként részesültek a DRFT-ban, nem származott előnyük belőle, sőt a térdextensió mértéke is csökkent. Így a DRFT ennél a csoportnál nem ajánlott. Esetleg második ülésben elvégezhető, ha szükséges, a fémek kivétele során. Összességében a SEMLS a járási deformitások kezelésére a cerebral paretikus gyerekek estében hatékony és javasolt technika.

9. Literature

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10. Publications

10.1. Publications directly related to the thesis

10.1.1. Full articles

1) Dreher T., **Vegvari D.**, Wolf S., Geisbüsch A., Simone Gantz, Wenz W. and Braatz F.: Development of knee function following hamstring lengthening in children with spastic diplegia – a long-term outcome study. *Journal of Bone and Joint Surgery- American Volume. 2012 Jan 18;94(2):121-30.* **(IF: 3.234)**
2) Dreher T, Wolf SI, Maier M, Hagmann S, **Vegvari D**, Gantz S, Heitzmann D, Wenz W, Braatz F: Long-term results after distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff-knee gait in spastic diplegic cerebral palsy. *Journal of Bone and Joint Surgery- American Volume. 2012 Oct 2;94(19):e 142(1-10)* **(IF: 3.234)**
3) Dreher T, **Vegvari D**, Wolf SI, Klotz M, Müller S, Metaxiotis D, Wenz W, Döderlein L, Braatz F: Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendineus lengthening in children with spastic diplegia. *Gait & Posture. 2013 Mar;37(3):430-5* **(IF: 2.299)**
4) **Vegvari D**, Wolf SI, Heitzman D, Klotz MC, Dreher T: Does proximal rectus femoris release influence kinematics in patients with cerebral palsy and stiff knee gait? *Clinical Orthopedics and Related Research. 2013 Oct;471(10):3293-300* **(IF: 2.882)**

10.2. Publications not directly related to the thesis

10.2.1. Full articles

1) Dreher T, Brunner R, **Vegvari D**, Heitzmann D, Gantz S, Maier MW., Braatz F, Wolf SI: The effects of muscle-tendon surgery on dynamic electromyographic patterns and muscle tone in children with cerebral palsy. *Gait & Posture. 2013 Jun;38(2):215-20* **(IF: 2.299)**

10.2.2. Abstracts

1) Szakály N, Tamás P, Terebessy T, **Végvári D**, Marschalkó P, Basch L: Internet database of scoliosis screening. *Biomechanica Hungarica VI: 2013 (1) 103-110*

2) Lukáts Á, Szabó A, Halász G, Berta ÁI, Röhlich P, Doma V, **Végvári D**, Szél Á: Photopigment coexpression in the mammalian retina. *XIX. International Symposium of Morphological sciences, Budapest, Hungary, 2007. augusztus 19-24./(Abstr) Acta Biol. Segediensis, 51 (suppl) 2007, 26*

3) Doma V, Halász G, Szabó A, Berta ÁI, **Végvári D**, Röhlich P, Szél Á, Lukáts Á: The effect of thyroid hormone substitution on M/L-cone development in in vitro organotypic retinal culture. *XIX. International Symposium of Morphological sciences, Budapest, Hungary, 2007. augusztus 19-24./ (Abstr) Acta Biol. Segediensis, 51 (suppl) 2007, 10*

4) **Végvári D**, Szabó A, Deák G, Lukáts Á, Berta ÁI, Szél Á: The expression of erythropoietin and its receptor in the developing rat retina. *XIX. International Symposium of Morphological sciences, Budapest, Hungary, 2007. augusztus 19-24./ (Abstr) Acta Biol. Segediensis, 51 (suppl) 2007, 52*

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CLINICAL RESEARCH

Does Proximal Rectus Femoris Release Influence Kinematics In Patients With Cerebral Palsy and Stiff Knee Gait?

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Abstract

Background Stiff gait resulting from rectus femoris dysfunction in cerebral palsy commonly is treated by distal rectus femoris transfer (DRFT), but varying outcomes have been reported. Proximal rectus femoris release was found to be less effective compared with DRFT. No study to our knowledge has investigated the effects of the combination of both procedures on gait.

Questions/purposes We sought to determine whether an additional proximal rectus release affects knee and pelvic kinematics when done in combination with DRFT; specifically, we sought to compare outcomes using the (1) range of knee flexion in swing phase, (2) knee flexion velocity and (3) peak knee flexion in swing phase, and (4) spatiotemporal parameters between patients treated with DRFT, with or without proximal rectus release. Furthermore the effects on (5) anterior pelvic tilt in both groups were compared.

Methods Twenty patients with spastic bilateral cerebral palsy treated with DRFT and proximal rectus femoris release were matched with 20 patients in whom only DRFT was performed. Standardized three-dimensional gait analysis was done before surgery, at 1 year after surgery, and at a mean of 9 years after surgery. Basic statistics were done to compare the outcome of both groups.

Results The peak knee flexion in swing was slightly increased in both groups 1 year after surgery, but was not different between groups. Although there was a slight but not significant decrease found the group with DRFT only, there was no significant difference at long-term followup between the groups. Timing of peak knee flexion, range of knee flexion, and knee flexion velocity improved significantly in both groups, and in both groups a slight deterioration was seen with time; there were no differences in these parameters between the groups at any point, however. There were no group differences in spatiotemporal parameters at any time. There were no significant differences in the long-term development of anterior pelvic tilt between the groups.

Conclusions The results of our study indicate that the short- and long-term influences of adding proximal rectus femoris release on the kinematic effects of DRFT and on pelvic tilt in children with cerebral palsy are negligible.

Level of Evidence Level III, therapeutic study. See the Guidelines for Authors for a complete description of levels of evidence.

Each author certifies that he or she, or a member of his or her immediate family, has no funding or commercial associations (eg, consultancies, stock ownership, equity interest, patent/licensing arrangements, etc) that might pose a conflict of interest in connection with the submitted article.
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Each author certifies that his or her institution approved the human protocol for this investigation, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

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Introduction

Biarticular muscle movement reduces energy consumption during walking because energy is transferred from one joint to the other [25, 37]. For this, a high level of motor control is needed to modulate muscle contraction (eccentric, concentric, or isometric) [14, 15]. Owing to this demand for a high level of motor control, biarticular muscle function is commonly disturbed and contractures may develop in children with cerebral palsy [17]. However, knowledge is limited regarding the synergism and connection between the proximal and distal parts of muscles in children with cerebral palsy, especially for the rectus femoris muscle.

In some children with cerebral palsy, activity is prolonged or continuous in the distal part of the rectus femoris muscle during the swing phase; this counteracts the knee flexion that is essential for an undisturbed clearance [1, 3, 8, 23, 30, 33]. This gait abnormality, known as stiff knee gait, causes increased energy expenditure and is accompanied by reduced gait velocity and step length and thus tripping and falling. Furthermore, electromyographic (EMG) activity is often abnormal in the rectus femoris muscle in the swing phase, while the vasti show a normal pattern [23, 33, 37]. A couple studies have shown that EMG activity of the rectus femoris in swing occurs too late for the muscle to decelerate the flexing knee given the delays between neural excitation and the development of muscle force [9, 10]. To improve knee flexion and swing phase clearance, standard surgical treatment involves distal femoris tendon transfer, which is commonly performed as part of single-event multilevel surgery. The beneficial effects after distal rectus femoris transfer (DRFT) for knee kinematics are well documented; however, inconsistent results have been reported [8, 11, 18, 20, 21, 27, 30, 33], and in some patients no effect was observed after DRFT [6]. Some authors suggested that even after transfer, a knee extensor moment remains in the rectus femoris [2, 3, 28].

The proximal part of the rectus femoris is believed to play an important role in pelvic tilt and hip flexion [16, 33]. To treat hip flexor contracture and increased anterior pelvic tilt, proximal rectus femoris release can be considered [16] and still is performed sometimes, although few studies have investigated its effects [16, 33]. However, McMulkin et al. [16] found no relevant influence of proximal rectus release on the knee in patients in whom DRFT was not performed [16]. Sutherland et al. [33] compared DRFT and proximal rectus release concerning the effect on knee kinematics and found significantly less effect for the group in which a proximal rectus release was performed.

The main effects of DRFT on stiff knee gait are seen mainly as consequences of its distal release and not as an augmentation of the knee flexor muscles [7]. Some authors reported recurrence of stiff knee gait in a relevant number of patients [6] and scarring at the transfer site is believed to be one possible reason for recurrence [7]. Fox et al. [7] reported a potential indirect effect of the hip flexion on the knee induced by the rectus femoris. In the literature, there are no reports on the outcome if both ends of the rectus femoris are surgically treated simultaneously.

The question therefore arises regarding whether an additional proximal rectus release affects knee function when done simultaneously with DRFT. The purpose of our study was to investigate the effects of an additional proximal rectus release on the knee kinematics when done in combination with DRFT. We sought to determine whether an additional proximal rectus release affects knee and pelvic kinematics when done in combination with DRFT; specifically, we sought to compare outcomes using the (1) range of knee flexion in swing phase (2) knee flexion velocity and (3) peak knee flexion in swing, and (4) spatiotemporal parameters between patients treated with DRFT, with or without proximal rectus release. The effects on (5) anterior pelvic tilt in both groups were compared.

Patients and Methods

For this retrospective study, all ambulatory (Gross Motor Functional Classification System Levels I to III [22]) patients with spastic bilateral cerebral palsy who were treated with DRFT in combination with proximal rectus femoris release as a part of multilevel surgery were selected from our gait laboratory database. Twenty patients were included (nine males and 11 females) with a mean age of 11 years at surgery. These patients had to have had a standardized preoperative examination and one short-term (1-year) and one long-term examination at least 5 years after surgery. In this study, the results of these patients were compared with those from patients who received only DRFT and not an additional proximal rectus femoris release, who also were selected from the database in a matched-pair analysis. The matching criteria included: maximum knee flexion in swing and knee flexion range in swing and age at surgery, BMI, and the Gillette Gait Index [31]. Anterior pelvic tilt at baseline was significantly higher in the group which received DRFT and rectus femoris release, indicating that increased anterior pelvic tilt was one criterion to perform proximal rectus release in this group. Furthermore a hip flexion contracture greater than 10° measured with the Thomas test [13] was a criterion for proximal rectus femoris release. The study was approved by the local ethics committee.

In the DRFT group (13 males and seven females), the mean age of patients at surgery was 9.9 years. Fifteen patients were independent walkers and five from each group needed walking devices (Table 1). All patients from

Table 1. Demographics, global walking ability, and surgical procedures

Variable	DRFT			DRFT and release		
	Preoperative	1 year postoperative	9 years postoperative	Preoperative	1 year postoperative	9 years postoperative
Followup (years)	–	0.91 (± 0.2)	8.47 (± 2.1)	–	1.00 (± 0.1)	9.17 (± 2.2)
Patient age (years)	9.94 (± 2.9)	11.05 (± 3.0)	18.31 (± 3.7)	11.00 (± 3.4)	12.29 (± 3.6)	20.58 (± 4.7)
BMI (kg/m^2)	16.83 (± 3.9)	17.77 (± 3.8)	20.66 (± 4.7)	18.11 (± 3.2)	19.29 (± 3.8)	21.77 (± 5.1)
Sex	7 F, 13 M			11 F, 9 M		
GMFCS Level I	2	3	5	1	3	4
GMFCS Level II	13	10	11	14	9	12
GMFCS Level III	5	7	4	5	8	4
Surgical procedures						
DRFT	40			40		
Proximal rectus femoris release	0			40		
Psoas over the brim	2			2		
Hamstring lengthening	32			28		
Calf muscle lengthening	32			38		
Femoral derotation osteotomy distal	14			8		
Femoral derotation osteotomy proximal	9			18		
Tibial derotation osteotomy	3			2		
Bony foot stabilization	15			12		
Soft tissue foot	12			6		
Total number of procedures	159			190		
Average number per patient	7.95			9.5		

DRFT = distal rectus femoris transfer; F = female; M = male; GMFCS = Gross Motor Function Classification Scale.

both groups had undergone preoperative, 1-year postoperative, and 9-year postoperative (8.5 years in the DRFT only group; 9.2 years in the DRFT and proximal rectus release group) computerized three-dimensional (3-D) gait analysis to determine the spatiotemporal parameters and kinematics. A conventional, marker-based Vicon® 3-D-motion capture system (Oxford Metrics, Oxford, UK) was used to track the 3-D positions of 25 reflective markers during walking according to the Plug-in Gait marker set (Oxford Metrics) [12]. The kinetic and kinematic data were collected simultaneously during level walking over a 7-m walkway. For the kinetic analyses, two floor-mounted force plates were used (Kistler®, Winterthur, Switzerland). All the data were integrated into a custom-made database. For each patient, 10 to 15 trials were recorded and the data of at least five representative strides of different trials were averaged. All examinations were performed by a specially trained physiotherapist (PA) and a study nurse (WS) with special education in neurodevelopmental disorders and long-term experience in working with children with cerebral palsy.

All surgical procedures were performed according to specific criteria based on clinical examination and gait analysis. All the patients had received single-event multilevel surgery, including soft tissue and bony operations (Table 1). The patients in the DRFT group received DRFT only, whereas the other patient group received DRFT and a proximal rectus femoris release. Standard surgical techniques were used for both procedures [6, 16, 38].

For data analysis, the preoperative, 1-year, and 9-year postoperative data of kinematic measurements, spatiotemporal parameters, Gillette Gait Index, and clinical examinations in both groups were compared using one-way repeated measures ANOVA (factor time) with Tukey's post hoc test (Prism 5; GraphPad Software, La Jolla, CA, USA) and t-test for group analyses. The level of significance was set at p less than 0.05.

Results

Knee ROM in swing significantly ($p < 0.05$) increased in both groups after surgery with no significant group differences ($p = 0.80$) (Table 2). Between the 1-year and the long-term examination, range of knee flexion in swing had deteriorated in both groups without any group differences ($p = 0.68$) (Fig. 1). However, this deterioration was only significant for

Table 2. Gait analysis outcome parameters

Parameters	Normal reference	DRFT			DRFT and release			Group difference
		Preoperative	1 year postoperative	9 years postoperative	Preoperative	1 year postoperative	9 years postoperative	
Kinematics								
Maximum pelvic tilt (°)	13 (5)	21 (7)	22 (7)	21 (9)	24 (8)	25 (7)	24 (6)	Preoperative (0.036)
Mean pelvic tilt (°)	12 (5)	16 (7)	19 (7)*	17 (9)	20 (7)	22 (7)	20 (7)	Preoperative (0.028); 1 year postoperative (0.049)
Maximum hip flexion in stance (°)	34 (6)	40 (11)	39 (8)	39 (9)	43 (9)	42 (10)	43 (10)	
Mean hip flexion in stance (°)	9 (6)	16 (10)	16 (10)	17 (8)	21 (9)	19 (9)	20 (9)	Preoperative (0.035)
Peak knee flexion in swing (°)	56 (6)	52 (12)	55 (8)	53 (10)	51 (10)	54 (9)	55 (8)	
Timing of peak knee flexion (% GC)	72 (1)	81 (4)	79 (5)*	78 (3)*	80 (5)	78 (5)*	78 (5)*	
Knee flexion velocity (°/% GC)	1.6 (0.2)	0.8 (0.4)	1.2 (0.4)*	1.0 (0.5)*#	0.8 (0.3)	1.2 (0.4)*	1.1 (0.4)*	
Knee ROM (°)	53 (5)	40 (15)	54 (13)*	45 (14)*#	38 (13)	52 (13)*	47 (13)*#	
Knee ROM in swing (°)	50 (5)	22 (8)	37 (10)*	34 (11)*	22 (8)	37 (10)*	33 (10)*#	
Minimum knee flexion in stance (°)	4 (4)	13 (19)	2 (13)*	8 (13)	13 (14)	1 (13)*	8 (11)#	
Spatiotemporal								
Timing of toe-off (% GC)	60 (1)	66 (5)	68 (6)	64 (4)*#	64 (5)	67 (6)*	64 (6)#	
Normalized cadence† (steps/ minute)	–	43.4 (7.5)	40.0 (9.1)	44.4 (6.6)	44.7 (11.1)	40.7 (11.3)	47.7 (10.3)	
Normalized speed† (m/second)	–	0.3 (0.1)	0.3 (0.1)	0.4 (0.1)#	0.3 (0.1)	0.3 (0.1)	0.4 (0.1)	
Normalized stride length† (m)	–	0.3 (0.1)	0.3 (0.1)	0.4 (0.1)*#	0.3 (0.1)	0.3 (0.1)	0.4 (0.1)	
Global gait								
Gillette Gait Index	–	426 (541)	212 (194)*	193 (116)*	350 (240)	187 (160)*	199 (198)*	

DRFT = distal rectus femoris transfer; GC = gait cycle; * significant difference from preoperative; #significant difference from 1 year postoperative; †this parameter is normalized to body height.

the group in which DRFT and proximal release was done. Knee flexion velocity improved significantly between baseline and 1 year postoperative in both groups (no group difference, $p = 0.55$) (Fig. 2). Between the 1- and the 9-year followups, a decrease in knee flexion velocity was found in both groups, but without any group differences ($p = 0.40$). Peak knee flexion in swing increased by approximately 3° in both groups (no group difference, $p = 0.98$) between the baseline and 1-year postsurgical examination. Although a slight decrease was found in the DRFT group, there was a slight increase in the group in which DRFT was done with additional proximal release between the 1-year and the long-term examinations. However, these changes were not statistically significant nor was there a significant group difference ($p = 0.30$). The timing of peak knee flexion decreased significantly after surgery in both groups (no group difference, $p = 0.43$) and remained without any group difference ($p = 0.98$) up to the long-term followup (Fig. 1). The minimum knee flexion (peak knee extension) in stance improved significantly in both groups after surgery (no group difference, $p = 0.75$). A significant recurrence in knee flexion in stance was found at the long-term examination for both groups without any group differences ($p = 0.91$) (Fig. 2).

The decrease in normalized cadence was not significant in both groups 1 year after surgery without any group differences ($p = 0.40$). At long-term followup the group with DRFT and release tended to show a higher normalized cadence ($p = 0.06$). The normalized speed in both groups initially decreased slightly 1 year after surgery without any difference between the two groups ($p = 0.37$). At long-term followup, normalized speed was found to be increased compared with 1 year postoperative in both groups (Table 2), however this increase was significant only for the DRFT only group ($p = 0.07$). The normalized stride length in the group with DRFT only was significantly ($p = 0.03$) improved at long-term followup, whereas the increase in the group with DRFT and release was not significant ($p = 0.13$). However, there was no significant difference in normalized stride length between the two groups at long-term followup ($p = 0.15$).

The Gillette Gait Index improved significantly in both groups ($p < 0.001$) in both groups. No significant differences were observed after surgery between the two groups at any examination (Table 2).

There was an increase of the pelvic anterior tilt between baseline and the 1-year postsurgical examination in both groups; this increase was significant in the DRFT only group and this group difference was significant ($p = 0.04$). In both groups between the 1- and the 9-year followups anterior pelvic tilt changed, and at long-term followup nearly baseline status was reached (Fig. 1). There was no significant group difference found at long-term followup ($p = 0.10$), indicating that function was comparable in both groups.

Discussion

An injury of the high-level motor control in patients with cerebral palsy affects the mechanisms of biarticular muscles considerably [24–26, 36]. The rectus femoris muscle, which was found to act independently of the vasti [19], is one of the biarticular muscles that is commonly involved in patients with cerebral palsy and causes stiff knee gait by reducing knee flexion during the swing phase of gait. It is believed to cause or aggravate increased anterior pelvic tilt. DRFT is commonly performed to correct stiff knee gait, however variable outcomes have been reported [5, 18, 20, 30]. A proximal rectus femoris release was done to eliminate rectus function at the pelvic level and to influence knee motion but was found to be less effective than DRFT. However, to our knowledge, the outcome after a combination of both procedures has not been reported. In our long-term study we could not find any beneficial effect of a proximal rectus femoris release on knee and pelvic kinematics and the indication for this procedure in addition to DRFT should be questioned.

Our study had numerous limitations. First, this was a retrospective study, which may influence the comparability of the two groups chosen, especially concerning the preoperative indication for selected surgical procedures. Furthermore, the patients in our two study groups had several additional procedures (eight procedures per subject on average; Table 1) as part of single-event multilevel surgery. Some concomitant procedures affect knee kinematics, such as hamstring lengthening, which improves knee motion in the long term [1, 5]. Hamstring lengthening may increase anterior pelvic tilt [5], since the hamstrings stabilize the pelvis and lengthening may lead to insufficiency. However, hamstring lengthening was done in a nearly equal number of patients in both groups. There were more patients who received a proximal femoral osteotomy in the group with additional rectus release compared with the group with DRFT only. This may have influenced the results. Because proximal rectus femoris release also was done to reduce increased anterior pelvic tilt, the group with additional rectus femoris release had a significantly higher ($p = 0.03$) mean pelvic tilt at baseline compared with the DRFT only group. This may bias a comparison of pelvic tilt development between the groups. In this study only sagittal plane kinematics were evaluated. Potential changes in frontal and transversal plane kinematics are not reported. Finally, gait analysis systems are subject to measurement errors owing to inherent system characteristics and marker placement variability.

Ounpuu et al. [20] and Sutherland et al. [32] reported that proximal rectus femoris release reduces hip flexion contracture and lumbar lordosis but also improves knee flexion in swing. Since it was shown that DRFT leads to a

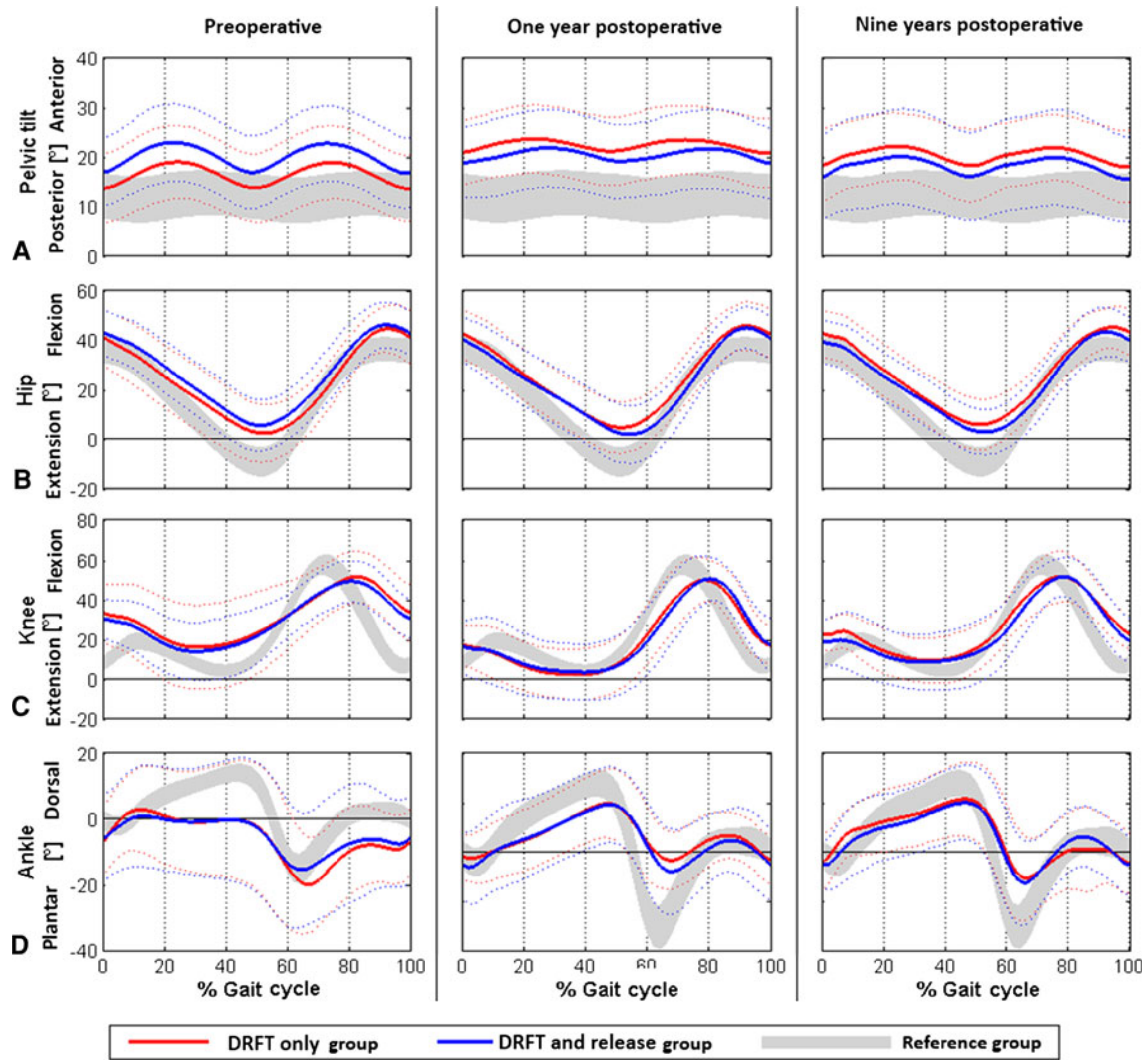
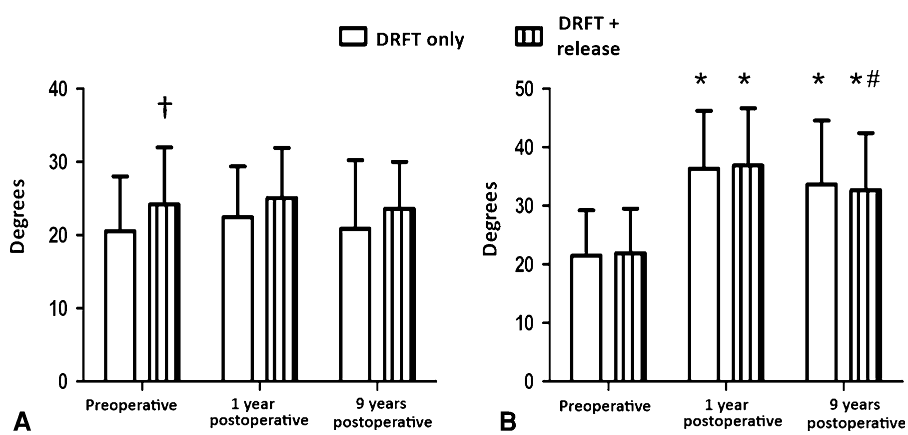


Fig. 1A–D Average sagittal plane kinematic mass graphs are shown for (**A**) pelvic tilt, (**B**) hip flexion, (**C**) knee flexion, and (**D**) ankle dorsiflexion and plantar flexion for all patients in the DRFT only group (red line), the DRFT and release group (blue line), and for an age-matched group of 48 subjects (gray area) (including 1 SD). The dotted red and blue lines correspond to ± 1 SD. Graphs are presented for all preoperative, 1-year postoperative, and 9- year postoperative examinations. Positive values indicate anterior pelvic tilt, hip flexion, knee flexion, and ankle dorsiflexion.

Fig. 2A–B The graphs show the mean and SD for (**A**) maximum pelvic tilt before surgery, 1 year after surgery, and 9 years after surgery, and (**B**) knee ROM in swing before surgery, 1 year after surgery, and 9 years after surgery. * Significant difference from preoperative; #significant difference from 1 year postoperative; †differences between DRFT group versus DRFT and release group.



superior outcome compared with proximal release, it has become the gold standard. There are numerous reports regarding the treatment of stiff knee gait using DRFT or proximal rectus femoris release [1, 4, 6, 8, 11, 18, 20, 21, 23, 27, 30, 32, 33, 35, 38]. Good initial results were reported [1, 4, 8, 11, 20, 21, 23, 27, 32, 33, 38]. However, some authors have reported discrepant outcomes after DRFT [6, 18, 30, 35], and a poor or no response rate of approximately 20% was found in a recent long-term investigation [6]. These inconsistent outcomes may be explained by a persistent extensor moment of the rectus femoris after transfer [2, 3, 28]. In such cases an additional proximal rectus femoris release may have an additional beneficial effect on knee motion.

According to our findings, the kinematic knee parameters (range of flexion in swing phase, peak knee flexion in swing, and knee flexion velocity) were significantly improved in both groups independent of whether rectus release was performed. The short-term improvements were comparable to those reported in the literature [18, 20, 21, 33, 35]. The effects in the DRFT only group of our study were sustained at the long-term followup, with the expectation of knee ROM in swing corroborating the findings of previous studies investigating long-term results after rectus femoris transfer [6, 18, 30, 35]. Knee ROM in swing decreased significantly in both groups but mainly as an indirect effect of recurrence in the stance phase knee flexion. When comparing the outcome of the DRFT only group with the group that received the additional proximal rectus femoris release, we could not find any group differences 9 years after surgery. Therefore, the results of our study indicate that the short- and long-term influences of proximal rectus femoris release on DRFT effects on the knee are negligible.

Two possible interpretations of our findings should be considered: (1) if DRFT effectively removes the knee-extending function of the rectus femoris, no surgery on the proximal part should directly influence knee kinematics. In this case our results would underline the effectiveness of DRFT in correcting stiff knee gait by eliminating the rectus femoris muscle [1, 6, 8, 11, 18, 20, 21, 23, 27, 30, 33, 35, 38]. However, since the rectus femoris may secondarily influence knee motion by generating an internal hip flexion moment and a secondary external knee flexion moment [7], the proximal rectus release is potentially able to influence the effects of DRFT on the knee. Our results that there were no differences between those patients who received additional proximal release and those who did not may indicate that the proximal rectus femoris release does not relevantly influence this mechanism. (2) In cases of a persistent internal knee extensor moment of the rectus femoris on the knee after DRFT [2, 3, 28], additional surgery in the proximal part does not affect knee function. Therefore, although clearly combined in biomechanics, clinically hip flexion and knee extension appear to be independent functions of the rectus femoris.

Causes of increased pelvis tilt can be a shortened rectus femoris or iliopsoas or/and weak hip extensors and poor selective motor control of the gluteus muscles and high femoral anteversion. Proximal rectus femoris release is performed to improve hip extension and to reduce increased anterior pelvic tilt. There were significant group differences in the maximum and mean pelvic tilt before surgery, reflecting the indication for an additional proximal rectus release in this group. This group difference remained at the 1-year followup for mean pelvic tilt, but at long-term evaluation no significant difference between the two groups was found. The pelvic tilt increased slightly in both groups after surgery and this increase should be seen mainly as the result of concomitant hamstring lengthening in many patients in both groups [5]. Therefore, the proximal rectus release did not prevent anterior pelvic tilt from increasing, corroborating the findings of McMulkin et al. [16]. This is underlined by the hip kinematics with no significant changes in hip motion being observed in either group. This is in accordance with the findings of Sutherland et al. [33]. We believe that surgery on the proximal parts of the rectus femoris does not relevantly influence pelvic and hip kinematics. It is a relevant question why we performed only a few psoas lengthening procedures [34]. We attempted to avoid psoas lengthening so as not to cause propulsion problems. It was done only if we were unable to correct a hip flexion contracture intraoperatively during proximal rectus release. It seems that increased hip flexion resolves after the correction of distal problems and this is in accordance with the results of Rutz et al. [29]. However, this needs to be investigated further.

We evaluated the effects on the knee and pelvis when DRFT and proximal rectus femoris release are performed simultaneously. Our short- and long-term results suggest that a combination of the two procedures does not further improve the effects of DRFT on stiff knee gait. Furthermore, we found no superior outcome concerning anterior pelvic tilt. Any additional effects of a concomitant proximal rectus femoris release on the knee and pelvis therefore are negligible.

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Development of Knee Function After Hamstring Lengthening as a Part of Multilevel Surgery in Children with Spastic Diplegia

A Long-Term Outcome Study

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Background: Hamstring lengthening commonly is performed for the treatment of flexed knee gait in patients with spastic diplegic cerebral palsy. Satisfactory short-term results after hamstring lengthening have been demonstrated in various studies. However, evidence for the effectiveness of hamstring lengthening to correct flexed knee gait is scant because of small and inhomogeneous case series, different surgical techniques, and short follow-up.

Methods: The long-term results for thirty-nine patients with spastic diplegia and flexed knee gait who were managed with intramuscular hamstring lengthening as a part of multilevel surgery are presented. Standardized three-dimensional gait analyses and clinical examinations were performed for all patients preoperatively and at one, three, and six to twelve years postoperatively.

Results: Significant improvements in kinematic parameters and the popliteal angle were noted at short-term follow-up ($p < 0.01$), supporting the results of previous studies. Long-term results showed significant deterioration of minimum knee flexion in stance and the popliteal angle ($p < 0.01$), whereas the improvements in the Gross Motor Function Classification System and Gillette Gait Index were maintained. This recurrence of flexed knee gait is partial and measurable. Increased pelvic tilt was found in 49% of the limbs postoperatively, which may represent one factor leading to recurrence of flexed knee gait. Genu recurvatum was seen in eighteen patients (twenty-seven limbs; 35%) one year postoperatively, especially in the patients with a jump knee gait pattern preoperatively. At long-term follow-up, genu recurvatum resolved in many limbs, but 12% of the limbs showed residual genu recurvatum, indicating that overcorrection represents a problem following hamstring lengthening.

Conclusions: The results of the present study are crucial for the prognosis of knee function after hamstring lengthening as a part of multilevel surgery. Recurrence and possible overcorrection should be considered in treatment planning.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

Ambulatory children with spastic diplegic cerebral palsy and gait disturbances commonly are managed with single-event multilevel surgery, which combines different osseous and soft-tissue procedures to achieve correction in all planes and on all levels[1-7]. Increased knee flexion during the stance phase of gait (flexed knee gait) is one of the most common gait abnormalities in spastic diplegia[8]. Rodda et al. further classified flexed knee gait into four patterns[9]: true equinus, jump knee, apparent equinus, and crouch. Hamstring spasticity with resultant contracture has been identified as one of the main factors leading to a flexed knee gait[8,10-15]. Later investigations have shown that increased external tibial torsion,

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TABLE I Demographic Data and Functional Baseline Parameters*†

Parameter	E0	E1	E2	E3
Time of follow-up‡ *(yr)*		1.0 ± 0.2	3.1 ± 1.0	8.1 ± 1.8
Age‡ *(yr)*	10.2 ± 3.5	11.3 ± 3.5	13.5 ± 3.3	18.5 ± 4.5
Gross Motor Function Classification System *(no. of patients)*				
Level I	6	11	14	13
Level II	21	11	16	13
Level III	12	17	9	13
Preoperative sagittal gait pattern§ *(no. of limbs)*				
Group I (true equinus)	0			
Group II (jump knee)	33			
Group III (apparent equinus)	12			
Group IV (crouch)	33			

*There were thirteen female patients and twenty-six male patients in the present study. †E0 = preoperatively, E1 = one year postoperatively, E2 = two to four years (mean, three years) postoperatively, and E3 = six to twelve years (mean, eight years) postoperatively. ‡The values are given as the mean and the standard deviation. §Sagittal gait pattern classification according to the system of Rodda et al.[9], obtained from preoperative, instrumented, three-dimensional gait analysis.

instability of the foot, and quadriceps weakness can cause or aggravate flexed knee gait[16-20]. Lengthening of the hamstrings is widely considered to be the standard surgical procedure for the correction of increased knee flexion[21-25]. The hamstrings can be lengthened with use of an open or a percutaneous technique[24,26-28]. Satisfactory short-term results with improved knee extension during stance phase have been shown in different studies[22,24,27,29]. However, outcomes have been inconsistent;

TABLE II Number of Surgical Procedures Performed in Single-Event Multilevel Surgery*

Procedure	All (N = 78)	Medial Hamstring Lengthening Group (N = 60)	Combined Medial and Lateral Hamstring Lengthening Group (N = 18)
Medial hamstring lengthening	78	60	18
Lateral hamstring lengthening	18	0	18
Psoas over the brim	14	7	7
Proximal rectus femoris recession	36	30	6
Adductor longus recession	10	7	3
Distal rectus femoris transfer	71	56	15
Calf muscle lengthening	54	45	9
Soft-tissue procedures, foot (split or complete posterior tibial tendon transfer, split anterior tibial tendon transfer)	18	14	4
Femoral derotation osteotomy	48	38	10
Tibial derotation osteotomy	5	2	3
Osseous foot stabilization (Evans calcaneal lengthening osteotomy; Grice extra-articular subtalar arthrodesis; Chopart or triple arthrodesis; calcaneocuboid joint distraction arthrodesis)	41	31	10

*The values are given as the number of limbs.

TABLE III Mean Values and Statistical Results for Selected Parameters*†‡

Parameter	E0	E1	E2	E3
Popliteal angle *(deg)*				
All	51 ± 18#**	30 ± 19§††	38 ± 16§††	49 ± 16#**
Medial hamstring lengthening	47 ± 17#	28 ± 19§††	37 ± 15††	51 ± 15#**
Combined medial and lateral hamstring lengthening	66 ± 11#	38 ± 17§	43 ± 19	43 ± 21
Speed *(m/s)*				
All	0.82 ± 0.23#	0.70 ± 0.32§**††	0.87 ± 0.30#	0.92 ± 0.30#
Medial hamstring lengthening	0.83 ± 0.23#††	0.70 ± 0.30§**††	0.88 ± 0.28	0.96 ± 0.28#
Combined medial and lateral hamstring lengthening	0.78 ± 0.21	0.71 ± 0.41	0.84 ± 0.37	0.79 ± 0.35
Cadence *(steps/min)*				
All	120 ± 24#	100 ± 30§**††	113 ± 26	119 ± 21
Medial hamstring lengthening	123 ± 26#	104 ± 27§	115 ± 25	114 ± 20
Combined medial and lateral hamstring lengthening	113 ± 13	88 ± 37	104 ± 27	104 ± 23
Stride length *(m)*				
All	0.82 ± 0.19**††	0.82 ± 0.24**††	0.91 ± 0.23§#	0.97 ± 0.21§#
Medial hamstring lengthening	0.82 ± 0.21††	0.79 ± 0.23**††	0.90 ± 0.23#††	0.99 ± 0.20§#**
Combined medial and lateral hamstring lengthening	0.82 ± 0.12	0.91 ± 0.23	0.95 ± 0.26	0.89 ± 0.24
Gillette Gait Index				
All	378 ± 240#**††	225 ± 172§	207 ± 168§	217 ± 134§
Medial hamstring lengthening	377 ± 267#**††	207 ± 142§	201 ± 177§	208 ± 130§
Combined medial and lateral hamstring lengthening	380 ± 129**	287 ± 249	228 ± 139§	246 ± 149
Mean pelvic tilt *(deg)*				
All	17 ± 9	21 ± 8**	17 ± 8#	18 ± 8
Medial hamstring lengthening	17 ± 8	20 ± 7**	17 ± 8#	18 ± 7
Combined medial and lateral hamstring lengthening	16 ± 10	22 ± 10	17 ± 10	20 ± 10
Knee flexion at initial contact *(deg)*				
All	38 ± 17#**††	17 ± 11§**††	22 ± 11§#	23 ± 10§#
Medial hamstring lengthening	36 ± 17#**††	16 ± 12§**††	22 ± 11§#	23 ± 10§#
Combined medial and lateral hamstring lengthening	45 ± 14#**††	19 ± 8§	20 ± 12§	23 ± 10§
Mean knee flexion in stance *(deg)*				
All	30 ± 19#**††	11 ± 12§**††	18 ± 13§#	20 ± 11§#
Medial hamstring lengthening	27 ± 18#**	10 ± 13§**††	17 ± 12§#	20 ± 11#
Combined medial and lateral hamstring lengthening	42 ± 20#††	15 ± 12§	20 ± 16	21 ± 14§
Minimum knee flexion in stance *(deg)*				
All	21 ± 22#**††	1 ± 14§**††	10 ± 13§#	12 ± 13§#
Medial hamstring lengthening	17 ± 20#**	0 ± 14§**††	9 ± 12§#	12 ± 12#
Combined medial and lateral hamstring lengthening	35 ± 23#††	7 ± 12§	12 ± 16	12 ± 16§
Maximum internal knee flexor moment *(Nm/kg*m)*	0.55 ± 0.27	0.40 ± 0.21	0.52 ± 0.27	0.55 ± 0.24
Maximum internal knee extensor moment *(Nm/kg*m)*	−0.37 ± 0.25	−0.47 ± 0.29	−0.42 ± 0.22	−0.50 ± 0.31

*The values are given as the mean and the standard deviation. The study included thirty-nine patients overall, with thirty patients in the isolated medial hamstring lengthening group and nine in the combined medial and lateral hamstring lengthening group. All outcome parameters were evaluated for all thirty-nine patients, with the exception of maximum internal knee flexor and extensor moment (which was evaluated for nineteen patients). †Significance was determined with repeated measures of analysis of variance with Bonferroni correction. The level of significance was set at $p < 0.05$. ‡E0 = preoperatively, E1 = one year postoperatively, E2 = two to four years (mean, three years) postoperatively, and E3 = six to twelve years (mean, eight years) postoperatively. §Significantly different from E0. #Significantly different from E1. **Significantly different from E2. ††Significantly different from E3.

many patients have had improvement, whereas other patients have had only little benefit or even worsening[24,26-30]. After short-term follow-up, some authors have reported increased pelvic tilt and a high prevalence of genu recurvatum as a consequence of overcorrection[23,24,27]. Evidence for the effectiveness of hamstring lengthening to correct flexed knee gait in spastic diplegia is scant because of small and inhomogeneous case series, different surgical techniques, and short follow-up. As a result, hamstring lengthening currently is viewed as controversial. A major problem is that, to our knowledge, there have been no long-term studies investigating the effects of hamstring lengthening in skeletally mature patients who were managed in childhood. Therefore, the long-term results in adolescents and adults who had had hamstring lengthening as a part of multilevel surgery in childhood were investigated in the present study. Our hypothesis was that correction of flexed knee gait seen at short-term follow-up is not maintained in the long term.

Materials and Methods

Standardized three-dimensional gait analysis and clinical examination were routinely performed for all ambulatory patients with cerebral palsy before and after surgery at our institution. Regular follow-up examinations were planned for each patient.

For the present study, all ambulatory diplegic patients with flexed knee gait who were managed with hamstring lengthening as part of multilevel surgery in childhood between 1996 and 2003 were selected from the gait laboratory database. One hundred and fifty-five patients were identified. For inclusion in the present study, the patients had to have undergone at least two standardized postoperative examinations (three-dimensional gait analysis and clinical examination) at one year and at two to four years after surgery. Ninety-four patients matched these criteria. All ninety-four patients were scanned for exclusion criteria and afterward were reinvited for a third long-term follow-up examination at least six years after surgery. The exclusion criteria were previous orthopaedic surgery (eight patients), dyskinetic cerebral palsy (five patients), botulinum-toxin-A injections in the six months prior to single-event multilevel surgery (five patients), severe mental retardation (three patients), and loss of walking ability (two patients). Three patients who had had revision surgery for hamstring lengthening or other secondary procedures between the examinations, which potentially may have influenced the results, were also excluded from the present study. After the exclusion of patients, sixty-eight patients remained eligible for reevaluation. Thirty-nine patients who had had a mean age (and standard deviation) of 10.3 ± 3.5 years (range, six to sixteen years) at the time of surgery could be reevaluated and were included in the present study. Subjects and/or families or caretakers gave informed consent to participate in the present study, which was approved by an institutional ethics committee.

Static findings, including knee range of motion and the popliteal angle[31], and dynamic findings, obtained by means of conventional, instrumented, three-dimensional gait analysis[32], were evaluated four times: preoperatively (E0) and at one year (E1), two to four years (E2), and six to twelve years (E3) postoperatively. Demographic data, the results of Gross Motor Function Classification System testing[33,34], and baseline sagittal pattern[9] distribution are displayed in Table I.

Hamstring lengthening was considered for patients who had increased knee flexion at initial contact (at least 20°) and/or in midstance (at least 10°) as observed with three-dimensional gait analysis in combination with at least 30° of knee extension deficiency on the popliteal angle test and/or any knee contracture.

All patients underwent standardized single-event multilevel surgery, including osseous and soft-tissue procedures (Table II). The intraoperative goal of hamstring lengthening for the correction was to achieve a popliteal angle of 20°. Medial hamstring lengthening was done first, through a 3 to 4-cm medial incision at the transition between the proximal and the distal half of the thigh, about 7 to 12 cm above the popliteal crease with the patient in the supine position. Fractional lengthening of the semimembranosus muscle was achieved by means of transverse incisions in the aponeurosis (aponeurotic lengthening) at a single level or at several levels if needed. The semitendinosus muscle was lengthened by means of intramuscular tenotomy on the same level through the same approach. The gracilis tendon frequently was used for distal rectus femoris transfer. If the correction was still insufficient (>20° on the popliteal angle test) after medial hamstring lengthening, the biceps femoris muscle was lengthened with use of a fractional intramuscular aponeurotic lengthening technique at the distal dorsolateral part of the thigh.

TABLE IV Adverse Effects*†

	E1	E2	E3
Genu recurvatum‡			
All (n = 78)	27	10§	9§
Medial hamstring lengthening (n = 60)	21	7	7
Combined medial and lateral hamstring lengthening (n = 18)	6	3	2
Group II (jump knee) (n = 33)#	15	8	8
Group III (apparent equinus) (n = 33)#	7	2	1
Group IV (crouch) (n = 12)#	5	0	0
Increased pelvic tilt**			
All (n = 78)	38	30§	36§
Medial hamstring lengthening (n = 60)	26	22	24
Combined medial and lateral hamstring lengthening (n = 18)	12	8	12

*The total number of limbs showing genu recurvatum or increased pelvic tilt is shown for all groups and examinations. †E1 = one year postoperatively, E2 = two to four years (mean, three years) postoperatively, and E3 = six to twelve years (mean, eight years) postoperatively. ‡Genu recurvatum is defined as >5° of extension in stance phase. §Significantly different from the previous time period (McNemar test). #Sagittal gait pattern classification according to Rodda et al.[9] obtained from preoperative, instrumented, three-dimensional gait analysis. **Increased pelvic tilt is defined as an increase of mean pelvic tilt of >5° in comparison with E0.

Postoperative management consisted of early mobilization with immediate weight-bearing. Epidural analgesia was used to manage postoperative pain during mobilization. In patients with additional osseous procedures, weight-bearing was done three to four weeks after surgery. In cases with residual knee contracture, controlled serial thigh casting was used postoperatively to achieve full knee extension within five to seven days. All patients were fitted with thigh night splints for six months to maintain knee extension.

To further describe the type of flexed knee gait, the patterns were classified for all patients according to the system of Rodda et al.[9] (Table I).

Significant correlations between the parameters of the left and right limbs were found ($p < 0.05$). Due to this dependence, data analysis was done only for the left limbs of the patients in selected parameters from the clinical examination and three-dimensional gait analysis. Furthermore, subgroup analysis was performed for the medial hamstring lengthening and combined medial and lateral hamstring lengthening groups. Descriptive statistics were used for basic statistical analysis. To show time-changing effects, one-way repeated-measures analyses of variance were applied. To assess significant changes in the prevalence of adverse effects (genu recurvatum, increased pelvic tilt) at the three different postsurgical examinations, the McNemar test was applied. The level of significance was set at $p < 0.05$, and the Bonferroni correction was employed to adjust for multiple comparisons. Statistical analysis was performed with use of PASW Statistics 18 (SPSS, Chicago, Illinois).

Source of Funding

No external source of funding was used for this investigation.

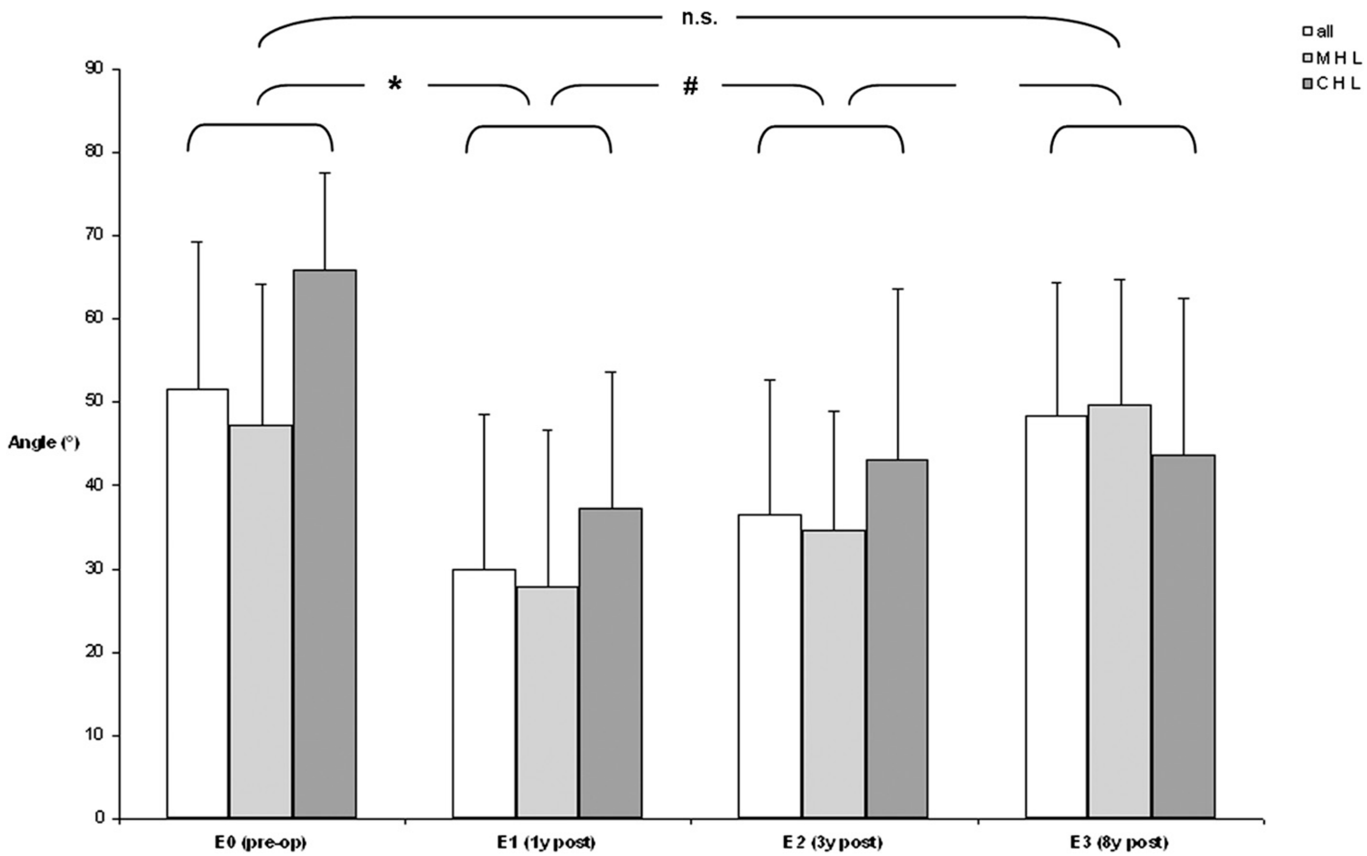

Fig. 1

Histogram showing the development of the popliteal angle (given as the mean and the standard deviation) at E0 (preoperatively), E1 (one year postoperatively), E2 (two to four years [mean, three years] postoperatively), and E3 (six to twelve years [mean, eight years] postoperatively) for all patients (n = 39), for the medial hamstring lengthening (MHL) group (n = 30 patients), and the combined medial and lateral hamstring lengthening (CHL) group (n = 9 patients). Positive values indicate knee flexion.*Significant difference between E0 and E1 ($p < 0.001$). #Significant difference between E1 and E2 ($p < 0.01$). ‡Significant difference between E2 and E3 ($p < 0.01$). n.s. = not significant.

Results

Clinical Examination and Walking Ability

The total number of independent walkers did not change over the years, whereas the Gross Motor Function Classification System levels improved, with seven more patients showing Gross Motor Function Classification System level I at E3 in comparison with E0 (Table I). No significant differences were observed in the Gross Motor Function Classification System level between the medial hamstring lengthening and the combined medial and lateral hamstring lengthening groups at baseline (E0).

A significant reduction in the mean popliteal angle was noted at E1 (Table III, Fig. 1). Between E1 and E3, all patients had significant deterioration of the popliteal angle. The popliteal angle deteriorated mainly between E2 and E3 in both groups. At E3, the popliteal angle was not significantly different from the preoperative values in either group.

Three-Dimensional Gait Analysis

Kinematics

The patients in both groups walked more slowly initially after surgery, and a significant increase in walking speed was seen only from E1 to E3 in the medial hamstring lengthening group. Cadence initially decreased after surgery in both groups but increased in the following years. Stride length increased significantly between E1 and E3 when both groups were considered together. However, when the groups were considered separately, the increase in stride length was only significant for the medial hamstring lengthening group.

The Gillette Gait Index was reduced significantly one year after surgery. The improvements in the Gillette Gait Index were maintained at the time of the long-term follow-up, and a significant difference in the Gillette Gait Index was shown between the long-term follow-up and baseline evaluations (Table III).

An increased mean pelvic tilt was found one year after surgery for both groups, particularly in the combined medial and lateral hamstring lengthening group (Table III). Pelvic tilt significantly decreased between E1 and E2 but showed another tendency for increase at the time of the long-term follow-up.

A significant reduction in knee flexion at initial contact, minimum knee flexion in stance, and mean knee flexion in stance was present in both groups initially after surgery (Table

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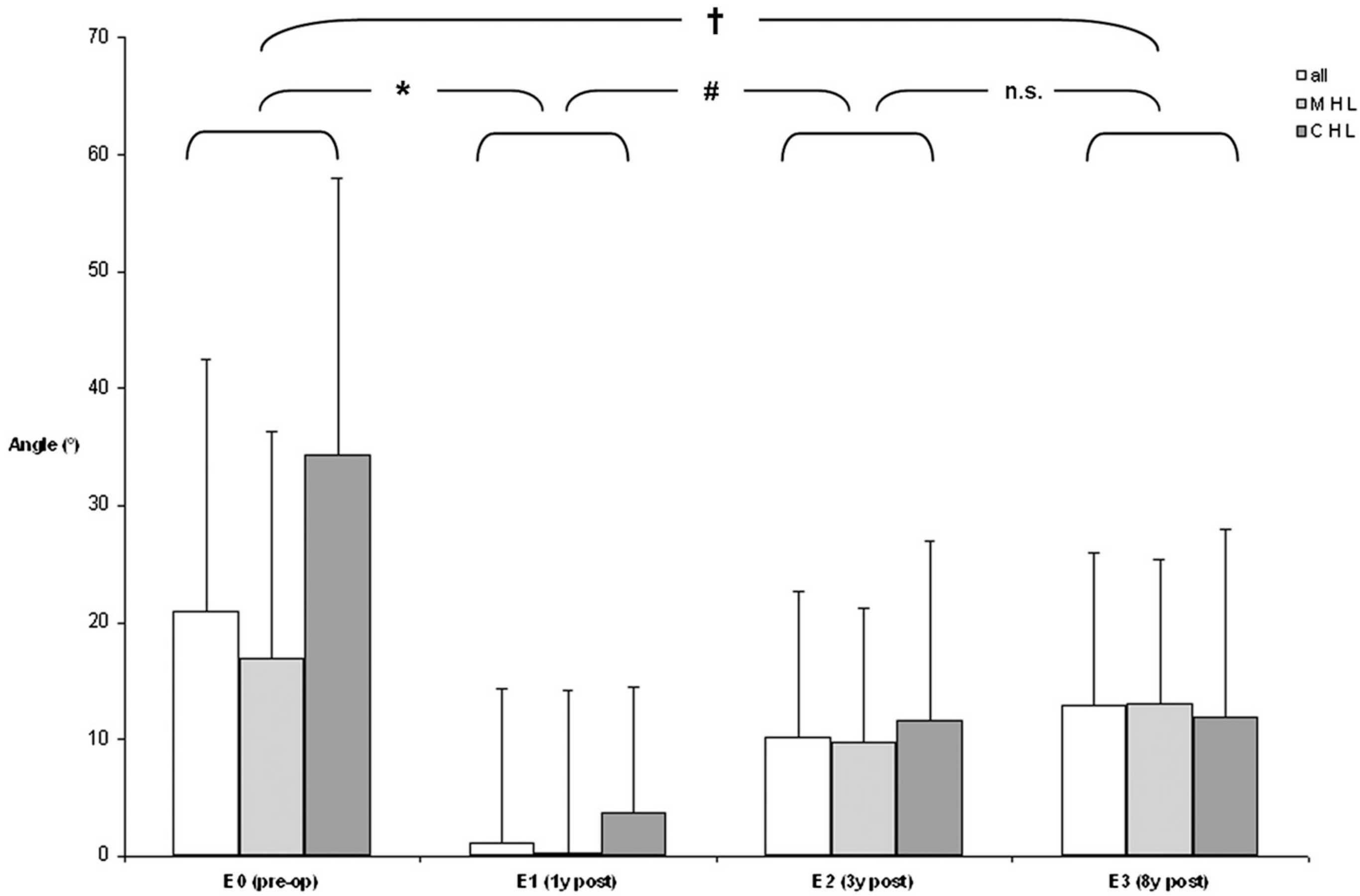

Fig. 2

Histogram showing the development of minimum knee flexion in stance (given as the mean and the standard deviation) at E0 (preoperatively), E1 (one year postoperatively), E2 (two to four years [mean, three years] postoperatively), and E3 (six to twelve years [mean, eight years] postoperatively) for all patients (n = 39), for the medial hamstring lengthening (MHL) group (n = 30), and for the combined medial and lateral hamstring lengthening (CHL) group (n = 9). Positive values indicate knee flexion. *Significant difference between E0 and E1 ($p < 0.001$). †Significant difference between E0 and E3 ($p < 0.01$). #Significant difference between E1 and E2 ($p < 0.05$). n.s. = not significant.

III). Minimum knee flexion in stance was reduced significantly by 17° for the medial hamstring lengthening group and by 28° for the combined medial and lateral hamstring lengthening group with respect to reference values. Over the years, a significant deterioration was found in all three knee kinematic parameters for all patients in comparison with E1 (Table III, Fig. 2), with 9° of deterioration noted for the minimum knee flexion in stance. Most of the deterioration was noted during the first three years after surgery. While the medial hamstring lengthening group showed a significant recurrence of minimum knee flexion in stance, the deterioration in the combined medial and lateral hamstring lengthening group was not significant. Comparison of long-term outcome and baseline values for the knee kinematic parameters in all patients showed that a significant improvement still could be found for the combined medial and lateral hamstring lengthening group but not for the less-involved medial hamstring lengthening group.

The average sagittal plane kinematics of the pelvis, hip, knee, and ankle at baseline, E1, E2, and E3 are shown in Fig. 3. The reference data for physiologic gait (twenty-four age-matched normalized patients) also are presented.

Kinetics

Kinetic data could not be recorded for patients who used a walking device but were recorded for the nineteen patients who were able to walk freely at all four examination dates. The maximum internal knee flexion moment for these nineteen patients was reduced at E1. This moment was recovered between E1 and E2 and was maintained at E3. The maximum knee extensor moment increased over the years. These changes were not significant.

Adverse Effects

The number of limbs showing genu recurvatum was calculated (Table IV). Genu recurvatum was defined as knee hyperextension of >5° in stance phase. At E1, genu recurvatum was found in eighteen patients (twenty-seven limbs, 35%), with nine patients having bilateral involvement and nine patients

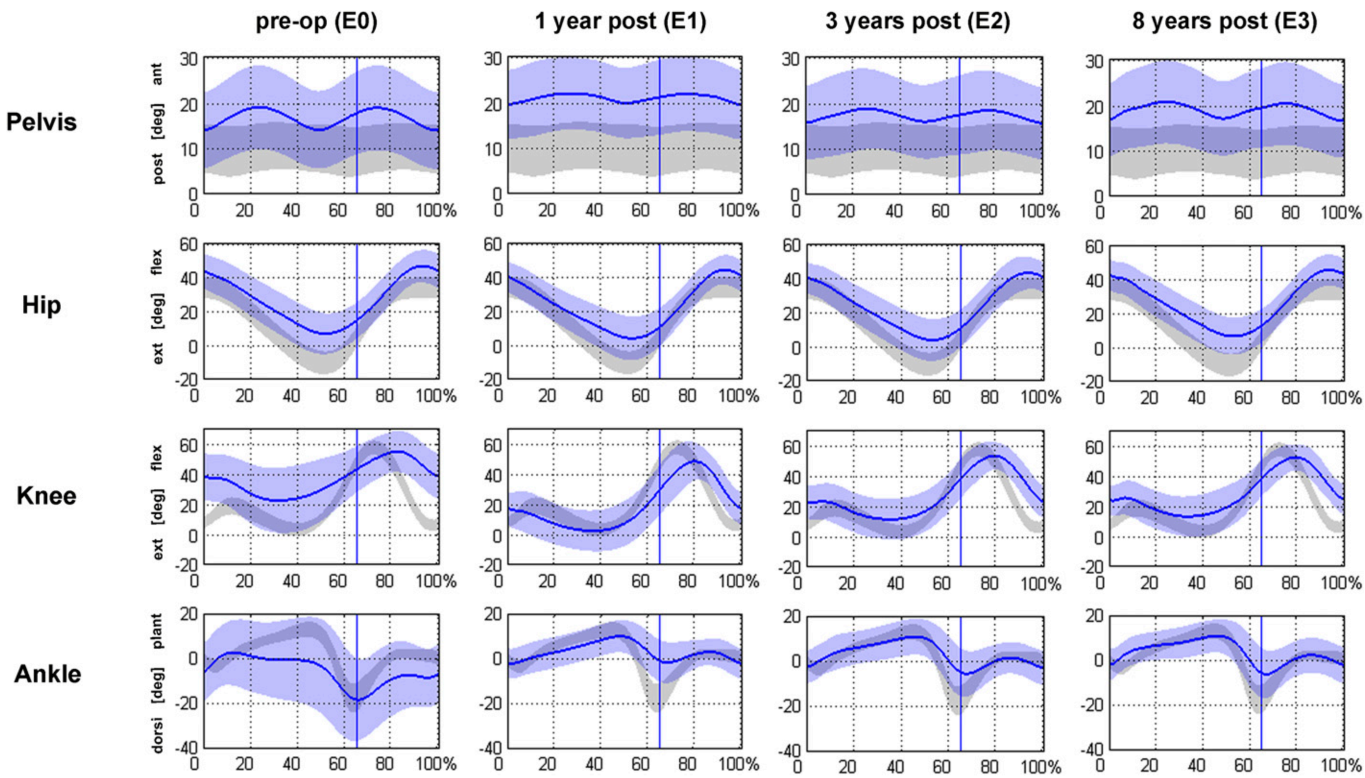

Fig. 3

Average sagittal plane kinematics as shown on gait analysis for all patients (n = 39) at E0 (preoperatively), E1 (one year postoperatively), E2 (two to four years [mean, three years] postoperatively), and E3 (six to twelve years [mean, eight years] postoperatively). The mean is represented by the blue line, the standard deviation is represented by the lucent blue area, and age-matched normalized data are represented by the gray area. Positive values indicate anterior pelvic tilt, hip flexion, knee flexion, and ankle dorsiflexion.

having unilateral involvement. At E2, the number decreased significantly to ten limbs (including six limbs in patients who had had bilateral involvement at E1 and four limbs in patients who had had unilateral involvement at E1). At the time of long-term follow-up, genu recurvatum was maintained in nine limbs (12%). No significant differences were found between the medial hamstring lengthening and combined medial and lateral hamstring lengthening groups in terms of genu recurvatum at any of the postoperative examinations. At E1, genu recurvatum occurred mainly in Groups II and III (representing 56% and 26% of the cases, respectively), with only 19% of the cases occurring in Group IV (the crouch group). At the time of long-term follow-up, there were no knee hyperextensions in Group IV, whereas residual genu recurvatum was observed in nine limbs in Groups II (eight) and III (one).

A clinically relevant increase in mean pelvic tilt (an increase of >5° in comparison with E0) was found in thirty-eight limbs (49%) at one year postoperatively (Table IV). The prevalence of increased pelvic tilt was 67% (twelve of eighteen) in the combined medial and lateral hamstring lengthening group, compared with 43% (twenty-six of sixty) in the isolated medial hamstring group. The overall prevalence of increased pelvic tilt decreased slightly between E1 and E2 to 38% (thirty of seventy-eight), but it increased again at the time of the long-term follow-up to 46% (thirty-six of seventy-eight).

Correlation Between Static and Dynamic Parameters

The correlation coefficient between the deterioration of the popliteal angle (between E1 and E3) and the recurrence of flexed knee gait on gait analysis (as represented by minimum knee flexion in stance) was calculated for all limbs. Only a weak correlation was found between the popliteal angle and minimum knee flexion in stance (Pearson correlation, $r_p = 0.29$; $p = 0.09$).

Discussion

Short-Term Outcome

Various short-term outcome studies have demonstrated improvement in terms of knee extension during stance phase and the popliteal angle following hamstring lengthening[21-30]. However, these short-term results have been inconsistent[22,24,26-30]. Comparable initial results in terms of the findings of gait analysis and the popliteal angle were found following intramuscular hamstring lengthening in the present study, corroborating these previous findings.

Long-Term Outcome

Because of central nervous system damage and growth, subsequent changes in gait pattern and joint function are expected after multilevel surgery. Investigations demonstrating results after more than three years of follow-up are rare. Adolfsen et al.

reported maintained improvement at four years after intramuscular hamstring lengthening in six patients with cerebral palsy[35]. In a four-year follow-up study after distal hamstring lengthening in ten children with cerebral palsy, Chang et al.[22] found an increase in the popliteal angle but no deterioration in kinematics. Gordon et al. reported maintained improvement in the popliteal angle and peak knee extension in stance at a mean of 2.8 years after medial hamstring tenotomy in a study of twenty-nine patients with cerebral palsy[27]. Rodda et al. reported maintained improvement in the popliteal angle and minimum knee flexion in stance in a five-year outcome study of ten patients with cerebral palsy[36]. The authors reported a slight but nonsignificant increase in minimum knee flexion in stance.

In the present study, outcome was evaluated an average of eight years (range, six to twelve years) postoperatively, and most patients were skeletally mature at the time of long-term follow-up because the average age at the time of surgery was ten years. The period between the ages of ten and eighteen years, in which further changes in gait may develop because of growth, is crucial for long-term outcome. In contrast to all of the intermediate-term studies discussed above, a significant deterioration of dynamic parameters, including mean knee flexion in stance, knee flexion at initial contact, and minimum knee flexion in stance, was found at long-term follow-up in the present study. Surprisingly, the improvements in the Gillette Gait Index and in the Gross Motor Function Classification System level were maintained, indicating that overall functional results after single-event multilevel surgery are preserved over the long term. However, both indices are global variables, which do not take into account detailed function of specific joints. Because the knee joint plays a central role in the gait of patients with cerebral palsy, the deterioration in knee kinematic parameters should be considered when planning the treatment of flexed knee gait. Additional lateral hamstring lengthening was needed in nine patients (eighteen limbs) to achieve adequate correction intraoperatively. Subgroup analysis showed that the medial hamstring lengthening and combined medial and lateral hamstring lengthening groups had identical sagittal knee parameters at long-term follow-up, but at the cost of increased pelvic tilt in the combined medial and lateral hamstring lengthening group.

The deterioration of the popliteal angle that was noted in the present study supports the findings of previous studies[37]. Thompson et al. proposed that the aim of flexed knee gait correction is primarily to improve function and not simply to improve static parameters[31]. The results of the present study underline that the deterioration of the popliteal angle and the recurrence of flexed knee gait as shown on three-dimensional gait analysis do not correlate well. As previously shown for other components of gait in patients with cerebral palsy, static clinical parameters are not useful as predictors of gait function[38-40]. The popliteal angle is not suitable as a method for predicting outcome or as a preoperative indication for hamstring lengthening.

Recurrence of Flexed Knee Gait

Various explanations for the recurrence of flexed knee gait should be considered as flexed knee gait represents a multiplanar problem and cannot be reduced to the knee joint only. Because of deterioration of the popliteal angle, recurrent hamstring tightness during growth represents one of the most frequently discussed possible causes. Furthermore, two different types of insufficiency may lead to recurrence over the years: proximal insufficiency (hamstrings) and distal insufficiency (lever arm dysfunction). Previous investigations have demonstrated the importance of muscle length to determine the dosage of lengthening and to avoid overcorrection[41-44]. Hamstring lengthening may lead to unwanted functional effects such as increased pelvic tilt (active insufficiency of the hamstrings). This may explain the first increase in pelvic tilt, seen one year postoperatively in the present study. Increased pelvic tilt may be compensated by increased lumbar lordosis or by walking with the knees in a flexed position (compensatory flexed knee gait), which may contribute to the recurrence. Further shortening of the hamstrings raises the pelvis, which may explain the improvements in pelvic position between E1 and E2. However, flexed knee gait will increase pelvic tilt again. Decreased foot lever arm, which is caused by instability of the foot or increased external or internal tibial torsion, also may cause increased knee flexion. Instability of the foot (midfoot break) frequently is seen in pes valgus, which commonly accompanies flexed knee gait[10]. Despite frequent osseous foot stabilization for the correction of pes valgus deformity in 53% of the limbs in the present study, flexed knee gait recurred.

Other reasons for a shortened foot lever arm are torsional deformities of the tibia[19,20] that were not treated during multilevel surgery. To our knowledge, no clinical studies have evaluated the importance of increased external tibial torsion or midfoot break with regard to the recurrence of flexed knee gait. In the present investigation, tibial malalignment was treated with supramalleolar derotation osteotomy in only five limbs at the time of multilevel surgery.

Weakness of the quadriceps muscle is another possible factor that may cause or aggravate flexed knee gait. In the present study, distal rectus femoris transfer was performed on seventy-one limbs (91%). The rectus femoris muscle contributes about 12% of the total quadriceps muscle power and mass[45], which potentially is lost when distal rectus femoris transfer is performed. Furthermore, many patients with cerebral palsy show continuous rectus femoris activity through the stance phase[46], which may lead to further weakness of the remaining vastii after distal rectus femoris transfer. Last, weight gain and increase in body mass index also may influence flexed knee gait and lead to recurrence[47].

Genu Recurvatum

Genu recurvatum is a common adverse effect of hamstring lengthening[22-24,27,35]. Adolfsen et al. identified genu recurvatum as a functionally relevant complication[35]. In the report by Kay et al., the prevalence of genu recurvatum was higher in the group that was managed with combined medial and lateral hamstring lengthening (16%)[24]. In the present study, genu recurvatum was found in 35% of the limbs, but there was no difference between the medial hamstring lengthening and combined medial and lateral hamstring lengthening groups. Knee hyperextension mainly occurred in patients with a preoperative jump knee gait pattern despite meticulous and successful equinus correction (Fig. 3, ankle kinematics), resulting in reduction of the excessive plantar flexion/knee extension couple. Therefore, this tendency for genu recurvatum seems to be attributed mainly to hamstring lengthening. Interestingly, the number of limbs with knee hyperextension decreased significantly, indicating that knee hyperextension may resolve during the years of follow-up. However, 12% of the limbs showed residual genu recurvatum at the time of the long-term examination, and all were in patients who had had a jump knee gait pattern preoperatively. Therefore, overcorrection represents a major complication, and a jump knee gait pattern should be taken into consideration before surgery. Another explanation for genu recurvatum at the time of long-term follow-up may be a recurrence of equinus deformity. The treatment of genu recurvatum is a challenge, and strategies are limited. In the present investigation, patients who had genu recurvatum postoperatively used an ankle-foot orthosis to prevent knee hyperextension when walking long distances.

Alternative Treatment

The proximal parts of the hamstring muscles are essential for hip extension and for stabilizing the pelvis[48]. The increased pelvic tilt in the present investigation supports the results of previous studies[22,23,41,49] and may represent one factor leading to recurrence. Alternative treatment strategies include the transfer of hamstring tendons to the femur to preserve the effects on the hip and pelvis[50-52]. Unfortunately, only a handful of investigations have evaluated the short-term results after hamstring transfer, and, to our knowledge, no long-term reports exist[50-52]. Another approach for correcting flexed knee gait is supracondylar femoral extension osteotomy used in combination with patellar tendon shortening in different investigations[53-58]. Encouraging short-term results have been reported[59,60]. Thus, the combination of femoral extension osteotomy and patellar tendon shortening represents our current treatment strategy. However, this approach cannot be recommended without reservation because long-term studies are lacking.

Limitations

There was a potential for selection bias in the present study that may limit the ability to generalize these results. In some locations children with cerebral palsy may not be seen for follow-up after the age of sixteen years. This is one possible reason for the general lack of long-term reports. In the present study, sixty-eight patients were eligible for reevaluation, of whom only thirty-nine (57%) returned for long-term follow-up.

Furthermore, the reported outcome may be influenced by other procedures that were performed at the time of multilevel surgery[61]. Therefore, the results of the present study should be interpreted as a cumulative effect of multilevel surgery, with hamstring lengthening representing one central procedure for knee function.

For the treatment of flexed knee gait, intramuscular hamstring lengthening as a part of multilevel surgery provides satisfactory short-term improvements in knee function during gait. However, the results of the present long-term study showed a significant recurrence of flexed knee gait. Increased pelvic tilt, found in approximately 50% of the limbs postoperatively, may be one factor leading to recurrence. Genu recurvatum, a possible consequence of overcorrection with hamstring lengthening, was seen frequently at one year postoperatively. Genu recurvatum may disappear over the years, but it remains a functional problem for patients with a preoperative jump knee gait pattern. Careful selection for surgery and appropriate hamstring lengthening are needed for those patients. Knowledge about the long-term effects and possible recurrence is of greatest interest when planning treatment and should be considered when providing preoperative information to patients and their parents. Extension osteotomy of the femur in combination with patellar advancement as a current alternative treatment strategy has led to encouraging short-term results[59,60], but it remains to be seen if these short-term results are maintained at long-term follow-up. ■

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Long-Term Results After Distal Rectus Femoris Transfer as a Part of Multilevel Surgery for the Correction of Stiff-Knee Gait in Spastic Diplegic Cerebral Palsy

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Background: The evidence for distal rectus femoris transfer as a part of multilevel surgery for the correction of stiff-knee gait in children with spastic diplegic cerebral palsy is limited because of inconsistent outcomes reported in various studies and the lack of long-term evaluations.

Methods: This study investigated the long-term results (mean, nine years) for fifty-three ambulatory patients with spastic diplegic cerebral palsy and stiff-knee gait treated with standardized distal rectus femoris transfer as a part of multilevel surgery. Standardized three-dimensional gait analysis and clinical examination were carried out before surgery and at one year and nine years after surgery. Patients with decreased peak knee flexion in swing phase who had distal rectus femoris transfer to correct the decreased peak knee flexion in swing phase (C-DRFT) were evaluated separately from those with normal or increased peak knee flexion in swing phase who had distal rectus femoris transfer done as a prophylactic procedure (P-DRFT).

Results: A significantly increased peak knee flexion in swing phase was found in the C-DRFT group one year after surgery, while a significant loss (15°) in peak knee flexion in swing phase was noted in the P-DRFT group. A slight but not significant increase in peak knee flexion in swing phase in both groups was noted at the time of the long-term follow-up. A significant improvement in timing of peak knee flexion in swing phase was only found for the C-DRFT group, and was maintained after nine years. Knee motion and knee flexion velocity were significantly increased in both groups and were maintained at long-term follow-up in the C-DRFT group, while the P-DRFT showed a deterioration of knee motion.

Conclusions: Distal rectus femoris transfer is an effective procedure to treat stiff-knee gait featuring decreased peak knee flexion in swing phase and leads to a long-lasting increase of peak knee flexion in swing phase nine years after surgery. Patients with more involvement showed a greater potential to benefit from distal rectus femoris transfer. However, 18% of the patients showed a permanently poor response and 15% developed recurrence. In patients with severe knee flexion who underwent a prophylactic distal rectus femoris transfer, a significant loss in peak knee flexion in swing phase was noted and thus a prophylactic distal rectus femoris transfer may not be indicated in these patients.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

Stiff-knee gait is a common gait disorder in patients with spastic diplegic cerebral palsy[1]. It is characterized by decreased knee flexion during swing phase, leading to foot clearance problems, reduced gait velocity, and reduced step length. Rodda et al.[2] classified stiff-knee gait as decreased knee excursion throughout the whole gait cycle of <30°, whereas

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Sutherland et al. described it as delayed and decreased peak knee flexion in swing phase with diminished total knee motion[3].

The primary cause of stiff-knee gait is the spasticity of the rectus femoris muscle[4-7], as it is pathologically active during swing phase, counteracting the knee flexion necessary for foot clearance and step length[4-8]. The standard surgical procedure for the treatment of stiff-knee gait is the distal rectus femoris transfer, which can be done either to the tendons of the medial or lateral hamstrings or to the iliotibial tract[5,7]. It aims to improve knee flexion in swing and peak knee flexion in swing for foot clearance. However, there are inconsistent criteria in the literature with regard to when a distal rectus femoris transfer should be performed[7-14].

Various studies have described good initial results, with an improvement in peak knee flexion in swing and knee flexion in swing following distal rectus femoris transfer[9-11]. Nevertheless, some authors have suggested that distal rectus femoris transfer does not generate a knee flexion moment, although the capacity of knee extension is diminished[12-14]. Other authors have noted no significant increase of peak knee flexion in swing but an increase of total knee motion[15-18]. Hence, the results after distal rectus femoris transfer are inconsistent. In addition, it is debatable whether patients with severe flexed knee gait who show highly increased knee flexion throughout stance phase and a normal or even increased peak knee flexion in swing phase should receive a prophylactic distal rectus femoris transfer to preserve peak knee flexion in swing after correction of flexed knee contractures by concomitant procedures such as hamstring lengthening or femoral extension osteotomy.

Only a few studies have addressed the longer-term outcomes after distal rectus femoris transfer. Saw et al. showed maintenance of improvement in peak knee flexion in swing phase, but the initial improvement in knee flexion in swing decreased and was associated with a loss of knee extension during stance phase 4.6 years postoperatively[9]. Moreau et al. also reported continuing benefits after a period of three years[10]. However, these studies were based on relatively small numbers, and both included patients with different types of cerebral palsy. Furthermore, recruitment was done from different treatment centers that used different surgical techniques of tendon transfer, and the time to the last follow-up was inconsistent, ranging from 0.9 to 6.7 years after surgery. Long-term studies with adequate patient numbers, homogeneous patient groups, and a follow-up interval of more than five years, when growth is expected to be finalized, are missing.

The purpose of the present study was the evaluation of long-term results (mean, nine years) in adolescents and adults with diplegic cerebral palsy who were treated with distal rectus femoris transfer as a part of single-event multilevel surgery in their childhood. Since the indication for distal rectus femoris transfer differed between patients with decreased peak knee flexion in swing and those with normal or increased peak knee flexion in swing, in which the distal rectus femoris transfer was done as a prophylactic procedure to preserve peak knee flexion in swing after the correction of severe flexed knee contracture in stance phase, these two groups were analyzed separately.

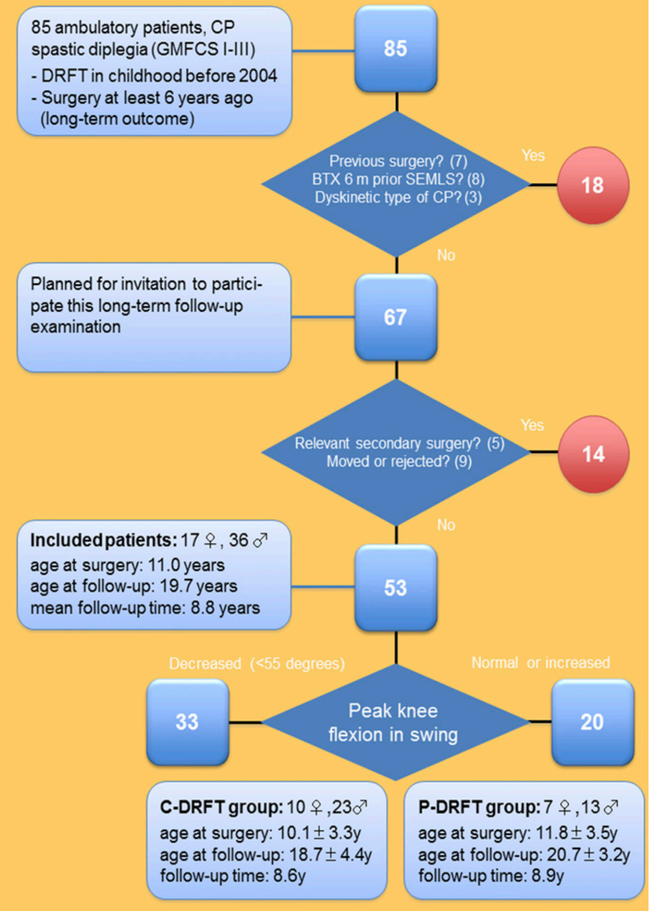

Fig. 1

Inclusion and exclusion criteria. Ages are given as the mean and the standard deviation. CP = cerebral palsy, DRFT = distal rectus femoris transfer, GMFCS = Gross Motor Function Classification System, BTX = botulinum toxin type-A injections, SEMLS = single-event multilevel surgery, C-DRFT = group that had distal rectus femoris transfer to correct decreased peak knee flexion in swing phase, and P-DRFT = group that had prophylactic distal rectus femoris transfer.

Materials and Methods

Ambulatory patients with cerebral palsy who are selected for surgical correction regularly receive a standardized evaluation, which includes conventional three-dimensional gait analysis and clinical examination, both before surgery and at regular follow-up examinations at our institution. Eighty-five ambulatory patients with spastic diplegic cerebral palsy (level I, II, or III, according to the Gross Motor Function Classification System [GMFCS][19,20]) who underwent distal rectus femoris transfer as a part of single-event multilevel surgery before 2004 were selected for this single-center cohort study from the gait laboratory database. Furthermore, these patients fulfilled the following inclusion criteria: an age of six to sixteen years at the time of surgery, a standardized follow-up examination at one year after surgery, and a positive preoperative Duncan-Ely sign[21,22]. Patients who received previous orthopaedic surgical treatment in the lower extremities (seven patients) or botulinum toxin type-A injections less than six months prior to single-event multilevel surgery (eight patients) and patients with dyskinetic type of cerebral palsy (three patients) were not included (Fig. 1). The remaining sixty-seven patients were invited for a long-term follow-up examination at least six years after intervention. Nine of these patients could not be evaluated because they had moved

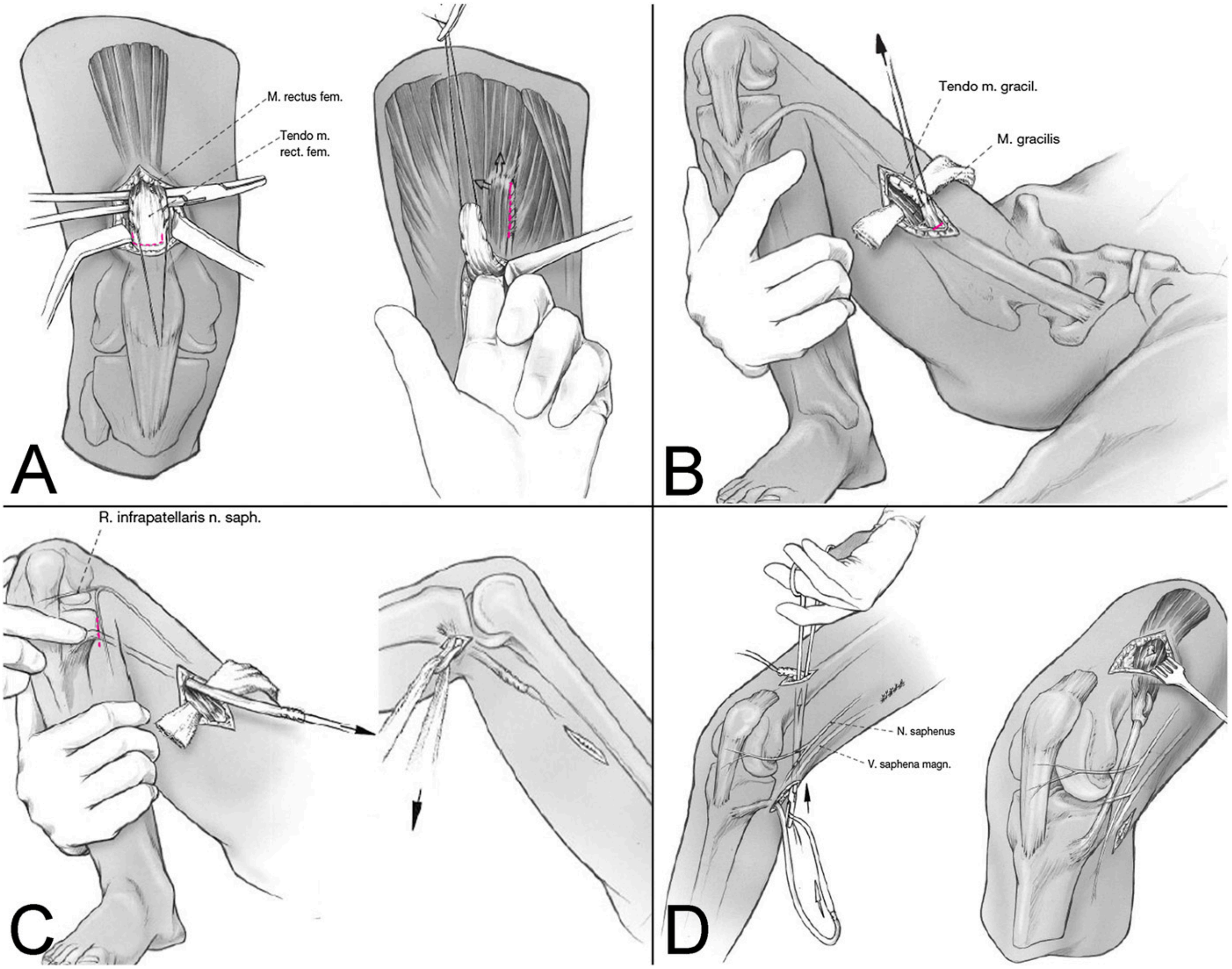

Fig. 2

The surgical technique of distal rectus femoris transfer. **Fig. 2-A** With the patient in the supine position, a 2-cm anterior approach is carried out at the distal part of the thigh, 3 to 4 cm above the proximal patellar pole. After incision of the fascia, the distal rectus tendon is identified and separated from the underlying remaining parts of quadriceps femoris. The tendon is tagged and released as distally as possible (red dotted line in left image). The rectus tendon is then mobilized and further separated from the remaining parts of the quadriceps femoris by digital preparation. Stronger adhesions are released by scissors or scalpel (red dotted line in right image). The fascia is dissected enough to avoid angular deviation of the transfer distally (red dotted line in right image). **Fig. 2-B** A medial thigh incision is made to isolate the gracilis muscle. The intramuscular tendon is exposed, tagged, and released proximally (red line) and is successively separated from its muscle belly distally while the knee is flexed. **Fig. 2-C** A mini-incision (red dotted line) at the posteromedial knee is used to expose and pull out the palpable distal gracilis tendon. **Fig. 2-D** Through the initial anterior approach, a long clamp is passed under the fascia and below the sartorius muscle belly to the mini-incision at the posteromedial aspect of the knee. The gracilis tendon is grasped by the tagging sutures and is transferred anteriorly, where it is sutured to the rectus femoris tendon with moderate tension while the knee is flexed 20°. (Reproduced, with modification, from Springer Science+Business Media: Wenz W, Döderlein L. Die Verpflanzung der Sehne des Musculus rectus femoris bei Patienten mit spastischer Diparese. Oper Orthop Traumatol. 1999;11(3):213-22. Reproduced with permission.)

(seven patients) or had declined to return for follow-up (one patient) or to participate in the study (one patient) because of personal reasons, and five patients were excluded because of relevant secondary surgery (one hip reconstruction surgery, two revision femoral derotation osteotomies, and two revision hamstring-lengthening procedures) (Fig. 1). Thus, fifty-three patients were evaluated at the long-term follow-up examination.

In total, fifty-three patients (seventeen female and thirty-six male with a mean age [and standard deviation] at the time of surgery of 11.0 ± 3.4 years) were re-evaluated at the long-term follow-up examination (mean age, 19.7 ± 3.8 years) (see Appendix). The subjects gave informed consent to participate in this study, which had been approved by the institutional ethics committee. Following a standardized protocol, the patients were examined before surgery and at one year and six to fourteen years after surgery. The mean long-term follow-up period was 8.8 ± 2.3 years after intervention. The evaluation included a standardized clinical examination and conventional three-dimensional gait analysis. Before 2002, examinations were carried out with use of a 120-Hz six-

TABLE I Clinical and Gait Analysis Outcome Parameters ➤

	C-DRFT Group*		
Parameters†	Preop. (E0)	One-Year Evaluation (E1)	Long-Term Evaluation (E2)
Clinical examination			
Duncan-Ely sign *(no. of patients)*	33§#	12#**	21§**
Passive knee extension†† *(deg)*	−3 (6)§#	1 (7)**	2 (6)**
Passive knee flexion†† *(deg)*	138 (15)	139 (12)	142 (6)
Three-dimensional gait analysis††			
Gillette Gait Index	361 (347) §#	136 (105)**	160 (121)**
Range of knee flexion in gait cycle *(deg)*	35 (11)§#	49 (12)**	47 (11)**
Range of knee flexion in stance *(deg)*	26 (9)§	33 (10)#**	29 (8)§
Range of knee flexion in swing *(deg)*	20 (8)§#	35 (10)**	34 (10)**
Knee flexion in stance *(deg)*	19 (10)§	11 (10)**	14 (10)
Knee flexion at initial contact *(deg)*	27 (9)§#	17 (9)**	18 (9)**
Minimum knee flexion in stance *(deg)*	10 (11)§	2 (12)**	5 (9)
Peak knee flexion in swing *(deg)*	45 (6)§#	51 (9)**	52 (9)**
Timing of peak knee flexion in swing *(% gait cycle)*	80 (6)§#	77 (4)**	77 (4)**
Knee flexion velocity *(deg/% gait cycle)*	0.8 (0.3)§#	1.2 (0.4)**	1.1 (0.4)**
Temporal parameters††			
Timing of toe-off *(% gait cycle)*	65 (5)	65 (4)	64 (4)
Step length *(m)*	0.4 (0.1)#	0.4 (0.1)#	0.5 (0.1)§**
Speed *(m/s)*	0.9 (0.3)#	0.9 (0.2)#	1.0 (0.3)§
Cadence *(steps/min)*	125 (24)§	115 (19)**	114 (21)

*The group that had distal rectus femoris transfer for correction of decreased peak knee flexion transfer (<55°) (C-DRFT) included sixty-six limbs in thirty-three patients, and the group with normal or increased peak knee flexion (≥55°) that had prophylactic distal rectus femoris transfer (P-DRFT) included forty limbs in twenty patients. †Analysis of variance (gait analysis parameters) and the McNemar-Test (Duncan-Ely test) with Bonferroni correction were used for statistical analysis of changes between examinations. The level of significance was a p value of <0.05. ‡The Levene and unpaired t tests were used to detect differences between groups at E0, E1, and E2, and Bonferroni correction was used. The level of significance was a p value of <0.05. §Significant difference compared with one-year postoperative results. #Significant difference compared with long-term results (mean, nine years). **Significant difference compared with preoperative findings. ††Values are given as the mean, with the standard deviation in parentheses, of the left legs.

camera Vicon 370 system (Oxford Metrics, Oxford, United Kingdom) and two force-plates (Kistler Instruments, Winterthur, Switzerland). Later, a 120-Hz twelve-camera Vicon 612 system (Oxford Metrics) was utilized, and equivalency of both systems was carefully checked. Skin-mounted markers were applied to osseous landmarks of the patients according to the Plugin Gait marker set (Oxford Metrics). Examinations were carried out by a specially trained physiotherapist and a study nurse specially experienced in pediatric neurodevelopmental therapy and gait analysis. Kinematics and kinetics were calculated according to the system described by Kadaba et al.[23]. Patients were asked to walk barefoot along a 7-m walkway at self-selected speed on both preoperative and postoperative analysis. At least five strides were averaged for further analysis.

Most patients underwent standardized distal rectus femoris transfer to the gracilis tendon (ninety-eight legs in forty-nine patients). When the gracilis tendon was too thin, distal rectus femoris transfer was done to the distal tendon of the semitendinosus muscle (eight legs in four patients). The surgical technique is illustrated in Figure 2. Concomitant surgical procedures performed during single-event multilevel surgery are given in the Appendix. Postoperative management included early mobilization with immediate weight-bearing transfers and subsequent walking within the first week after single-event multilevel surgery. Knee-ankle-foot orthoses in knee extension were applied at night for six months. Depending on concomitant osseous procedures, this schedule was modified accordingly.

Indication criteria for distal rectus femoris transfer generally included a positive Duncan-Ely sign on clinical examination, a decreased knee flexion in swing, and decreased peak knee flexion in swing seen in gait analysis as well as pathological increased activity of the rectus femoris muscle with dynamic electromyography[5-11,15-18]. In those patients, distal rectus femoris transfer was done to correct knee flexion in swing and peak knee flexion in swing (the C-DRFT group). In patients with severe flexed knee gait who demonstrated normal or increased peak knee flexion in swing, distal rectus femoris transfer was done as a prophylactic procedure (the P-DRFT group) to preserve peak knee flexion in swing and to improve knee flexion in swing after the correction of flexed knee in stance phase. These two groups were evaluated separately.

Statistical analysis of the data was done only for the left legs of the patients, as indicated by significant right-left correlations ($p < 0.05$). Limbs with peak knee flexion in swing phase that was one standard deviation below the age-matched reference value (<55° of peak knee flexion in swing) were assigned to the C-DRFT group (thirty-three limbs), while the other limbs were assigned to the P-DRFT group (twenty limbs) for further analysis (Fig. 1). Typical clinical and three-dimensional gait analysis parameters were used for data evaluation according to previous reports[9-11]. In addition, the Gillette Gait Index[24] (GGI) was calculated for all patients at all examinations. Normative data were obtained from an age-matched healthy reference group. Descriptive statistics were used for basic statistical analysis. To show the time-changing effects of each group, one-way repeated-measures analyses of variance (ANOVA)

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TABLE I (*continued*)

P-DRFT Group*			
Preop. (E0)	One-Year Evaluation (E1)	Long-Term Evaluation (E2)	Significant Differences Between Groups†
20§#	12#**	18§**	E1, E2
−13 (11)§#	−2 (7)**	−2 (8)**	E0, E2
136 (18)	139 (11)	133 (16)	
486 (282)#	319 (198)	290 (176)**	E1, E2
29 (15)§#	45 (14)**	40 (13)**	E2
19 (9)§#	34 (12)#**	26 (10)§**	E0
19 (8)§#	32 (10)**	29 (9)**	E2
45 (18)§#	16 (14)**	23 (12)**	E0, E2
49 (17)§#	21 (10)#**	27 (9)§**	E0, E2
38 (20)§#	7 (14)#**	15 (13)§**	E0, E2
67 (12)§#	52 (12)**	54 (7)**	E0
82 (5)	81 (6)	81 (5)	E1, E2
0.6 (0.3)§#	1.0 (0.4)**	0.9 (0.4)**	E2
67 (5)§	71 (9)**	68 (7)	E2
0.4 (0.1)	0.4 (0.1)	0.4 (0.1)	E2
0.7 (0.2)	0.6 (0.3)	0.7 (0.3)	E0, E1, E2
110 (26)§	82 (33)**	96 (24)	E0, E1, E2

were applied. For the evaluation of group differences, Levene and unpaired t tests were used. P values of <0.05 were considered significant, and Bonferroni correction was used to adjust for multiple comparisons. Pearson correlations were calculated between baseline parameters and their improvement between baseline and the time of the long-term follow-up. Statistical analysis was done using PASW Statistics 18 (SPSS, Chicago, Illinois).

Source of Funding
No external source of funding was used for this investigation.

Results

Three-Dimensional Gait Analysis

A significant increase of peak knee flexion in swing was found at one year for the C-DRFT group, while a significant decrease was found in the P-DRFT group. Between the one-year and long-term follow-up evaluations, peak knee flexion in swing tended to increase in both groups. Concerning the long-term outcome, peak knee flexion in swing remained significantly ($p < 0.01$) increased in the C-DRFT group, while it remained significantly decreased in P-DRFT group (Table I). The delay in timing of peak knee flexion in swing was significantly improved ($p < 0.01$) after distal rectus femoris transfer in the C-DRFT group at one year and was maintained at the time of the long-term follow-up ($p < 0.01$), whereas no significant changes were detected for the P-DRFT group.

The knee flexion in swing phase was significantly increased at one year in both groups ($p < 0.001$), whereas this increase was significantly higher in the C-DRFT group ($p < 0.01$). At the time of the long-term follow-up, this increase was maintained in the C-DRFT group, while a tendency for decrease ($p = 0.08$) was found in the P-DRFT group (Table I).

The stance phase parameters (mean knee flexion in stance, knee flexion at initial contact, and minimum knee flexion in stance) were significantly decreased from baseline in both groups ($p < 0.001$), although this decrease was significantly higher in the P-DRFT group because of the significantly greater knee flexion contracture at baseline in comparison with the C-DRFT group ($p < 0.01$). Both groups showed deterioration of these parameters between one year and long-term follow-up examination, although this deterioration was significantly higher in the P-DRFT group ($p < 0.01$).

A significant increase in knee flexion velocity was found in both groups at one year ($p < 0.01$). At the time of the long-term follow-up, there was a marginal decrease found in both groups, which was not significant, and the difference between the long-term follow-up and preoperative evaluations was still significant ($p < 0.01$) (Table I). The mean knee kinematic curves of the two groups and the age-matched reference group at all three examinations are visualized in Figure 3.

Temporal Parameters

Step length did not significantly increase between the preoperative and one-year evaluations in either group but increased significantly between the one-year and long-term follow-up examinations in the C-DRFT group ($p < 0.001$). This increase may be explained by the increase of leg length during growth. Walking speed was reduced in the P-DRFT

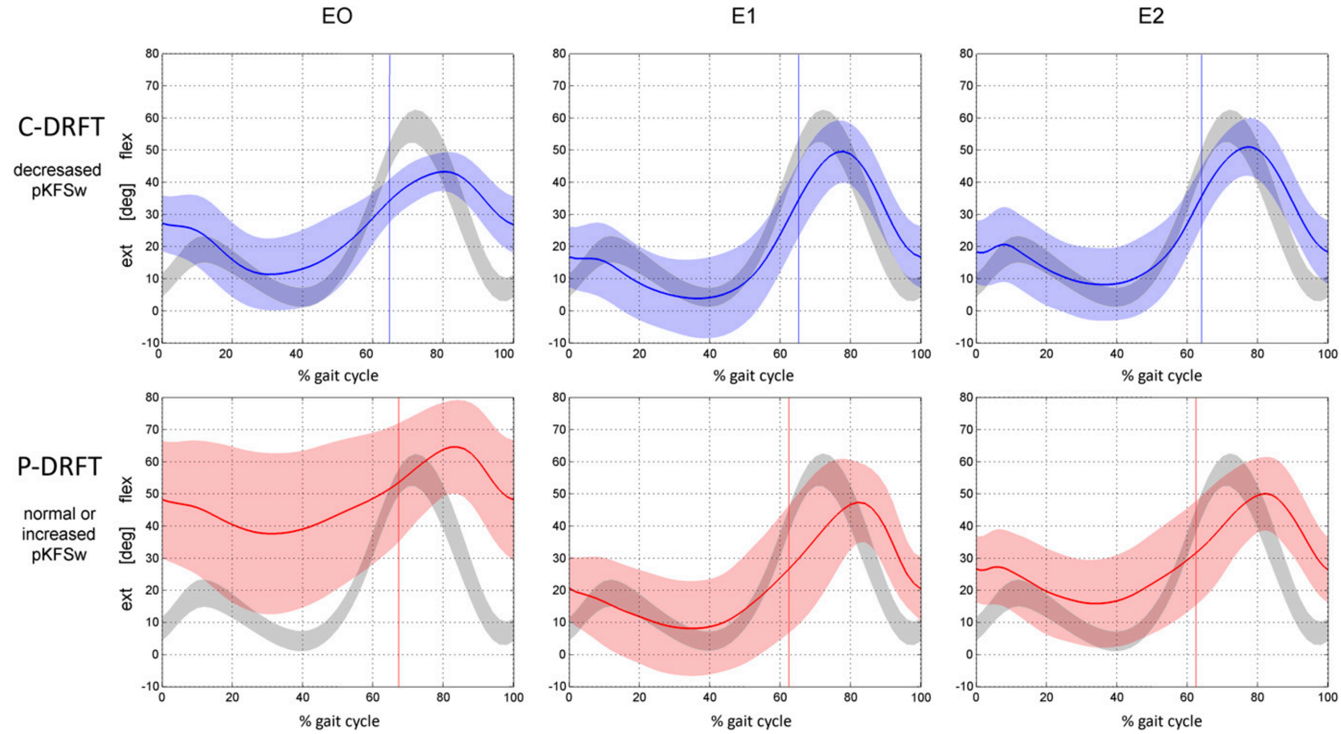

Fig. 3

Sagittal plane knee kinematics. Graphs are shown for the preoperative (E0; left column), one-year postoperative (E1; middle column), and nine-year postoperative (E2; right column) examinations. In the top row, the mean patient data for the C-DRFT group are represented by the blue line and the standard deviation is displayed as the lucent blue area. In the bottom row, the mean patient data for the P-DRFT group are represented by the red line and the standard deviation is displayed as the lucent red area. The reference data for physiologic knee kinematics obtained by an age-matched group of twenty-five normal subjects are also presented (gray area). Positive values indicate knee flexion. pKFSw = peak knee flexion in swing.

group initially after surgery but increased between the one-year and nine-year follow-up. A significant increase in walking speed was found for the C-DRFT group between baseline and long-term follow-up. Cadence was not significantly increased between the preoperative and long-term follow-up evaluations (Table I).

Global Gait Variables

The GGI was significantly improved between the preoperative and one-year evaluations and was maintained at the time of the long-term follow-up in both groups (Table I). The patients in the P-DRFT group had severe flexed knee contracture. Hence, their GGI level was significantly higher preoperatively ($p < 0.01$), and the overall improvement was smaller in comparison with that of the C-DRFT group.

In the C-DRFT group, three patients with GMFCS level II improved to level I, whereas four patients with level II needed walking devices after surgery (level III). Two more patients with level II preoperatively reached level I at the time of the long-term follow-up. Two patients with level III preoperatively had level II at the time of the long-term follow-up. Two patients with level II preoperatively needed walking devices at the long-term evaluation.

In the P-DRFT group, two patients with level II reached level I, four patients with level III reached level II, and two patients with level II were classified as level III at the time of the long-term follow-up.

Clinical Examination

Significantly fewer patients had a positive Duncan-Ely sign at one year compared with the number with a positive sign preoperatively ($p < 0.01$), especially in the C-DRFT group (Table I). However, between the one-year and long-term follow-up, the number of patients with positive Duncan-Ely sign increased significantly in both groups. This increase was significantly higher in the P-DRFT group ($p < 0.05$), and no significant difference was found between preoperative and long-term evaluations. Six limbs showed moderate knee flexion contracture between 10° and 20° preoperatively in the C-DRFT group. In the P-DRFT group, six limbs had moderate and ten had severe (between 20° and 40°) knee flexion contracture preoperatively.

Response and Recurrence

With regard to knee flexion in swing, a good response was defined as an increase of knee flexion in swing of at least 10° at one year postoperatively. Accordingly, an increase of <10° was classified as a poor response.

At one year, twenty-two limbs (67%) in the C-DRFT group and fifteen limbs (75%) in the P-DRFT group were

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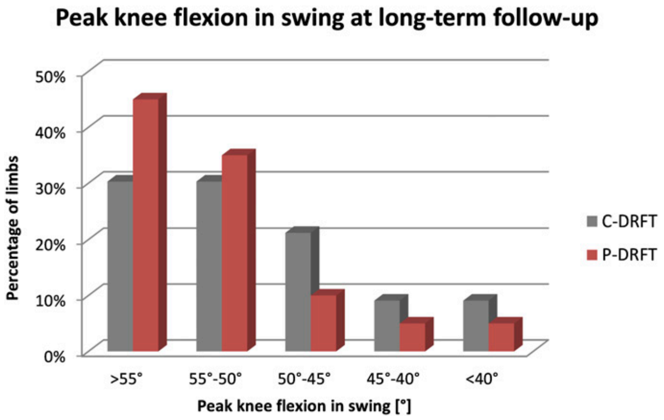

Fig. 4

The percentage of limbs in different categories of resulting peak knee flexion in swing at the time of the long-term follow-up in the group that had distal rectus femoris transfer for correction of decreased peak knee flexion (C-DRFT; gray bars) and the group with normal or increased peak knee flexion that had prophylactic distal rectus femoris transfer (P-DRFT; red bars). Thirty-nine percent of the limbs in the C-DRFT group and 20% of the limbs in the P-DRFT group presented with peak knee flexion in swing below two standard deviations (<50°) of the age-matched reference value at the time of the long-term follow-up.

classified as having a good response. Eleven limbs (33%) in the C-DRFT group and five limbs (25%) in the P-DRFT group showed a poor response. At long-term follow-up, the groups showed comparable rates of limbs with a good response (67% in the C-DRFT group and 65% in the P-DRFT group), whereas 15% and 5%, respectively, had a late response. Eighteen percent of the C-DRFT group and 15% of the P-DRFT group showed a permanently poor response. The preoperative and nine-year postoperative knee kinematics of a typical patient with a poor response are illustrated in the Appendix. Fifteen percent of the C-DRFT group and 20% of the P-DRFT group developed recurrence of a stiff-knee gait with regard to knee flexion in swing.

With regard to peak knee flexion in swing, 39% of the limbs in the C-DRFT group and 20% of the limbs in the P-DRFT group had peak knee flexion in swing below two standard deviations (<50°) of the age-matched reference value at the time of the long-term follow-up (Fig. 4). Distal rectus femoris transfer was not able to increase (C-DRFT group) or to maintain (P-DRFT group) peak knee flexion in swing in those patients.

Correlations

A significant correlation ($r_p = -0.8$; $p < 0.0001$) was found between the peak knee flexion in swing and the amount of its improvement (the difference between preoperative and long-term follow-up evaluations). A scatterplot is shown in Figure 5. The patients with more involvement have more potential to benefit from distal rectus femoris transfer. In patients with >53° of peak knee flexion in swing preoperatively, an increase of peak knee flexion in swing is not to be expected.

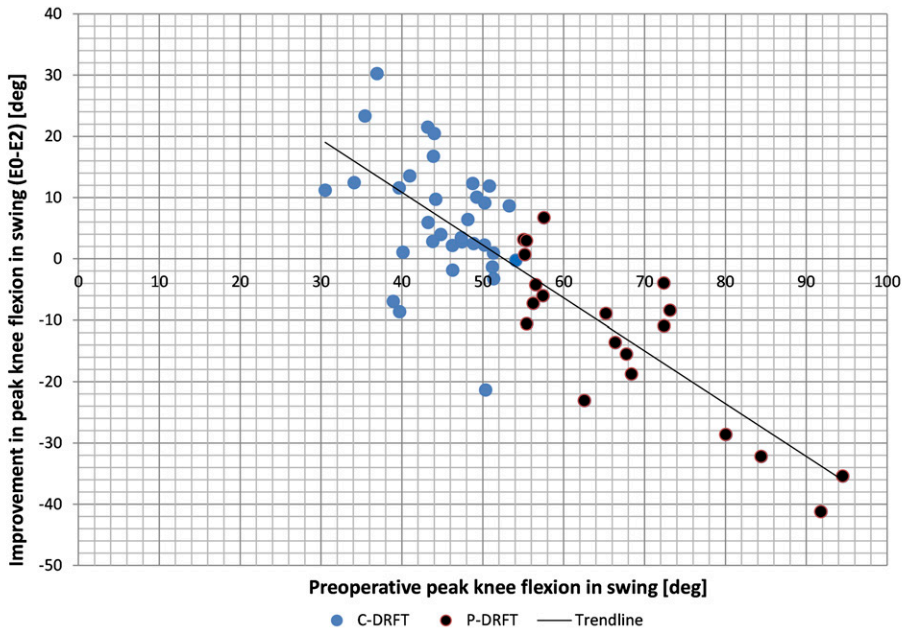

Fig. 5

Scatterplot for the preoperative status (x-axis) of peak knee flexion in swing (degrees) correlated with its improvement (y-axis; degrees), defined as the difference between the value at the time of the long-term follow-up and the baseline value. Each spot represents one limb. Limbs of the group that had distal rectus femoris transfer for correction of decreased peak knee flexion (C-DRFT) are blue, and those of the group with normal or increased peak knee flexion that had prophylactic distal rectus femoris transfer (P-DRFT) are shown in black. The trend line was drawn. The correlation was found to be significant (p < 0.0001). Limbs with more involvement showed more potential to benefit from distal rectus femoris transfer than limbs in which peak knee flexion was already high. A benefit for peak knee flexion in swing cannot be expected when the preoperative peak knee flexion is >53°.

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Discussion

The treatment of stiff-knee gait in cerebral palsy is challenging and controversial because variable effects following distal rectus femoris transfer have been reported[15-18,25-28]. Since natural progression in patients with cerebral palsy may lead to recurrence or aggravation of gait disturbances during growth[29,30], it is insufficient to consider only the short-term outcome. Only a few studies have previously investigated longer-term results following distal rectus femoris transfer. Saw et al. reported maintained improvements in swing phase flexion but a significant loss of knee motion due to progressive crouch gait 4.6 years after distal rectus femoris transfer in eighteen patients with cerebral palsy[9]. Limitations of that study were the small number of patients and the inclusion of different types of cerebral palsy. In a comparative study, Moreau et al. showed benefits after distal rectus femoris transfer in comparison with another cohort in which distal rectus femoris transfer was not carried out[10]. The positive effects of distal rectus femoris transfer were maintained in a second evaluation three years after surgery.

Our study is the first, to our knowledge, to separately evaluate patients with normal or increased peak knee flexion in swing (commonly those with severe flexed-knee contractures) who underwent distal rectus femoris transfer as a prophylactic procedure to prevent peak knee flexion in swing (the P-DRFT group) and patients with decreased peak knee flexion in swing, for whom the aim was a correction of decreased peak knee flexion in swing (the C-DRFT group). Mixing these two groups in previous reports may have masked improvements of peak knee flexion in swing after distal rectus femoris transfer[15-18]. In the present study, significant improvements in knee flexion in swing and knee flexion velocity one year after surgery were found for both groups, corroborating the results of previous investigations. However, peak knee flexion in swing and its timing were significantly improved in the C-DRFT group only, while peak knee flexion in swing was significantly decreased in the P-DRFT group and the timing did not change. The benefits in knee flexion in swing in the P-DRFT group seem to mostly occur because of the sizeable improvements of knee extension in stance phase. Despite prophylactic distal rectus femoris transfer in the P-DRFT group, there was a mean decrease of 15° in peak knee flexion in swing, which means that the entire curve was shifted downward. Prophylactic distal rectus femoris transfer was not able to maintain peak knee flexion in swing within one standard deviation of the norm, but a comparable average value was noted in comparison with the C-DRFT group. However, the question arises as to whether these effects can be attributed to the distal rectus femoris transfer or whether single-event multilevel surgery without prophylactic distal rectus femoris transfer would lead to the same results. In the patients with decreased peak knee flexion in swing preoperatively, the benefits in knee flexion in swing are mainly based on the mean improvement in peak knee flexion in swing (6°), which adds to the improved knee extension in stance phase, whereas the improvements in knee flexion in swing in the P-DRFT group are primarily the result of improved knee extension in stance. The increase of knee motion during swing phase in combination with an increased peak knee flexion in swing represents a benefit for foot clearance in swing phase and is of great relevance for a stable, undisturbed, and smooth gait as approximately 60° of knee flexion is necessary for regular swing foot clearance[7,31]. In the study by Hemo et al., conventional distal rectus femoris transfer to the medial hamstrings was compared with distal rectus femoris transfer attached to the iliotibial band, and no differences in outcome were found between the techniques[11]. The improvements found after distal rectus femoris transfer are mainly related to the elimination of the rectus femoris as a knee extensor and the prevention of reattachment of the distal rectus tendon in its anatomic anterior position by transferring it to another site.

Our long-term follow-up showed that the improvements in knee kinematics could be maintained and even enhanced nine years after surgery in the C-DRFT group. The overall good results nine years after surgery indicate that distal rectus femoris transfer carried out as a part of multilevel surgery is an effective approach for the treatment of stiff-knee gait. However, contrary to the benefits in the C-DRFT group, the knee flexion in swing in the P-DRFT group deteriorated. This may be partially explained by the recurrence of flexed knee gait, which was significantly higher in the P-DRFT group. Since the correction of stance phase extension is the primary goal in these patients, the approach of performing distal rectus femoris transfer as a prophylactic procedure is questioned because it weakens the knee extensors. This weakness may be one possible factor leading to recurrence of flexed knee gait in the P-DRFT group.

We showed that the patients with more involvement have more potential benefit from distal rectus femoris transfer. According to the scatterplot (Fig. 5), a benefit for peak knee flexion in swing cannot be expected when the preoperative peak knee flexion in swing is >53°. Some patients showed a poor response to distal rectus femoris transfer, and some developed recurrence of stiff knee gait. Different possible explanations should be considered. First, a persistent extensor moment may result from the transferred rectus muscle because of scar tissue or remaining extensor moment arm despite the transfer. This would underline the findings of Asakawa et al. and Riewald and Delp[32-35], who noted that the rectus femoris does not turn into a knee flexor and may have a persistent knee extensor moment. This would also explain the persistence of the Duncan-Ely sign in our study. Second, increased muscle tone of the remaining quadriceps muscle may lead to a poor outcome in some patients. Various studies have tried to outline a precise outcome prediction for the distal rectus femoris transfer[36-38]. In a recent study, Reinbolt et al. developed a model for outcome prediction in consideration of different gait analysis variables and found a prediction accuracy of up to 80%[39]. However, some uncertainty remains, potentially resulting in an unnecessary surgical procedure.

The question arises as to whether alternative treatment options such as distal release of the rectus femoris tendon are more effective. Sutherland et al. and Ounpuu et al. showed that rectus femoris release did not lead to a comparable or even better outcome in comparison with distal rectus femoris transfer[3,40]. In a recent study, Cruz et al. reported a comparable outcome after intramuscular distal rectus femoris recession and postulated that this procedure represents an alternative treatment strategy for the correction of stiff-knee gait[41]. However, peak knee flexion in swing and knee flexion in swing were not significantly improved. Recurrence after intramuscular lengthening is often seen in cerebral palsy[42], and recurrence of stiff gait has to be expected. The long-term effects of this technique are unclear.

There are some limitations of the present investigation. A potential for selection bias in this study may have influenced the results. There were nine patients who could not be evaluated because they had moved or they declined participation. A potentially bad outcome in these patients may have had an influence on the results. However, this is a well-known problem of long-term studies. This study is the first, to our knowledge, to describe the long-term outcome at a mean of nine years after distal rectus femoris transfer in patients who were nearly all skeletally mature adults. Another important factor that may influence the development of clinical and kinematic parameters during the years is growth and increase of the body mass index. These influences on outcome after multilevel surgery in general are not adequately addressed. Furthermore, concomitant procedures carried out during single-event multilevel surgery may have an influence on knee kinematics. Some patients received an additional proximal rectus femoris release for the correction of hip flexion contracture and double-bump pattern of the pelvis on gait analysis. The possible influence of proximal rectus release on knee kinematics, which has been reported to be limited[43], should be considered when interpreting the outcome in this study.

In conclusion, distal rectus femoris transfer leads to long-lasting improvements of peak knee flexion in swing, timing of peak knee flexion in swing, and range of knee flexion in swing in patients with preoperatively decreased peak knee flexion in swing, although patients with more involvement had the most improvement. Distal rectus femoris transfer as a part of multilevel surgery has the potential to counteract the natural progression of knee function limitation in patients with spastic diplegia. However, since 18% of the patients showed a permanently poor response, distal rectus femoris transfer has no influence in some patients, and different mechanisms may be responsible for recurrence, which was found in 15% of the patients. When patients with severe knee flexion contractures had prophylactic distal rectus femoris transfer, a significant loss in peak knee flexion in swing was noted. Single-event multilevel surgery without distal rectus femoris transfer may have resulted in the same outcome, and it is questionable whether distal rectus femoris transfer, which weakens knee extensor power, is indicated in these patients, for whom improvement of knee extension is the primary aim of treatment.

Appendix

eA Tables showing demographic data and global walking ability and the concomitant surgical procedures during single-event multilevel surgery as well as a figure demonstrating the sagittal knee kinematics of a patient with a poor response are available with the online version of this article as a data supplement at jbjs.org. ■

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Long-term effects after conversion of biarticular to monoarticular muscles compared with musculotendinous lengthening in children with spastic diplegia

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ABSTRACT

Adverse effects such as increased anterior pelvic tilt (APT) are reported after muscle-tendon lengthening (MTL) for the correction of flexed knee gait in cerebral palsy. The conversion of biarticular muscles (CBM) to monoarticular muscles represents an alternative treatment, but only few short-term results have been published, without comparison with MTL. The long-term outcome of 21 diplegic patients treated with CBM in a prospective study was compared with the results in MTL patients in a matched-pair analysis. Standardized clinical examination and three-dimensional gait analysis were done before surgery, 1 year thereafter, and at long-term follow-up a mean of 9.2 years postoperatively. Mean APT increased one year after surgery in both groups. This increase was higher in MTL patients and statistically significant only for this group. Knee flexion at initial contact and minimum knee flexion in stance were significantly decreased in both groups, while in swing the CBM group tended to show more of a decrease in knee flexion but at the cost of reduced peak flexion. Both groups showed deterioration of kinematic knee parameters through to long-term follow-up; the favourable effects of CBM disappeared, and the two groups displayed comparable average pelvic and knee kinematics. Considering individual patterns the prevalence of increased APT was lower in the CBM group 1 year after surgery, indicating that sparing the semitendinosus may have a positive effect on pelvic stability. However, after 9 years 30% of the patients in both groups showed increased APT indicative of persistent hamstring insufficiency. These results demonstrate that CBM, a significantly more extensive procedure, has no long-term advantage over MTL.

1. Introduction

Patients with diplegic cerebral palsy show a variety of gait disorders, of which flexed knee gait is one of the most frequent [1–3]. These gait problems are commonly treated by single-event multilevel surgery (SEMLS) [4–7]. Various surgical strategies have been employed for the correction of flexed knee gait. Lengthening of the hamstring muscles is widely accepted as the standard treatment [6,8–13]. Encouraging short- and mid-term results have been reported after hamstring lengthening [6,8–13]. However, a number of authors reported adverse effects after hamstring lengthening, such as genu recurvatum and increased anterior pelvic tilt (APT) [9–11,13]. The occurrence of these unintended effects can be partially explained by the influence of the lengthening of biarticular muscles on both adjacent joints [14]. Patients with cerebral palsy show more difficulties in controlling biarticular muscles than monoarticular muscles [15,16]. Elongation of the muscle-tendon unit by surgical means may add to muscle weakness after surgery [17]. As weakness of specific muscle groups due to primary brain damage is commonly found in patients with cerebral palsy, this surgical elongation aggravates the weakness of these muscles [18].

To avoid these adverse effects in the treatment of flexed knee gait, conversion of biarticular muscles to monoarticular muscles has been introduced [19–22]. In an early report, Silfverskiöld described conversion of the gastrocnemius, the hamstrings and the rectus femoris into monoarticular muscles [19,20]. Using a modified technique, Eggers transplanted the medial and lateral hamstring tendons to the femoral condyles [21]. Satisfactory initial results after transfer of the semitendinosus tendon to the adductor magnus tendon were published by Ma et al. [22]. Metaxiotis et al. found significant improvements after conversion of the semitendinosus and gastrocnemius muscles to monoarticular muscles [16].

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Interest in the conversion of biarticular to monoarticular muscles (CBM) has diminished, probably because of the lack of long-term reports comparing its results with those of conventional muscle tendon lengthening surgery (MTL) by means of three-dimensional gait analysis. The purpose of this study was therefore to evaluate the long-term effects of CBM in comparison with those of MTL. The hypothesis of the present investigation was that CBM and MTL lead to comparable outcomes concerning knee kinematics, but that CBM carries less risk than MTL for increased APT and recurrence of flexed knee gait.

2. Methods

2.1. Subjects

Before 1998, the standard approach for correction of flexed knee gait in spastic diplegia at our institution was MTL. Due to the frequent occurrence of increased APT and genu recurvatum, a prospective cohort study was conducted to evaluate the outcome of CBM. Between 1998 and 2004 one cohort of 25 diplegic children underwent CBM during SEMLS. These children met the following inclusion criteria: spastic diplegia, ambulatory (GMFCS I–III), age at surgery 6–16 years, flexed knee gait, scheduled for SEMLS. The exclusion criteria were athetosis, previous lower limb orthopaedic surgery, and botulinum toxin injections less than 6 months before surgery. The study was approved by the institutional ethics committee. All the subjects were evaluated according to a standardized protocol before operation and 1 year thereafter. The results were published [16]. For the present investigation, the same patients were invited to attend for a long-term follow-up examination at least 6 years after their multilevel surgery. Twenty-one of the 25 patients could be re-evaluated 6–14 years after surgery. The remaining patients had moved ($n = 2$), were not able to attend ($n = 1$) or preferred not to participate ($n = 1$). For the sake of comparison, the CBM patients were matched to 21 diplegic patients with flexed knee gait from our gait laboratory database who had received MTL during SEMLS. The patients were selected according to the following preoperative parameters: knee flexion and ankle dorsiflexion in stance (primary matching parameters), together with pelvic tilt, hip flexion, age at surgery, body mass index (BMI), Gillette Gait Index (GGI) and GMFCS level (secondary matching parameters). No significant differences in all these preoperative parameters were detected between the two groups (one-way ANOVA, $p < 0.05$). The selected MTL patients were evaluated following the same study protocol.

2.2. Examinations

Forty-two patients (21 CBM, 21 MTL) were examined before the intervention (E0) and 1 year (E1) (CBM: 1.3y ± 0.6y; MTL: 1.2y ± 0.7y) and 6–14 years (E2) (CBM: 9.2y ± 2.5y; MTL: 9.1y ± 2.6y) thereafter using the same protocol. Demographic data are summarized in Table 1. Standardized clinical examination including range of motion and special tests (Thomas test, popliteal angle, Silfverskiöld test) as well as instrumented three-dimensional gait analysis, carried out with a 50-Hz six-camera Vicon® 370 system (Oxford Metrics, Oxford, UK) and two force-plates (Kistler Instruments, Winterthur, Switzerland) until 2002. Subsequently a 120-Hz 12-camera Vicon® 612 system (Oxford Metrics) was used and the equivalence of the two systems was tested. Kinematics and kinetics were calculated according to Kabada et al. [23]. Each patient was asked to walk along a 7-m walkway barefoot at a self-selected speed, and at least five representative strides were averaged for further analysis.

2.3. Surgery

All patients received standardized SEMLS (Table 1). All procedures were performed according to specific clinical and gait analysis criteria under the supervision of one of the authors (L.D.). Treatment of the two groups differed with regard to the following biarticular muscles: semitendinosus, gastrocnemius and rectus femoris. In the CBM group, the semitendinosus tendon was released from its insertion with the patient in prone position. The medial tendon origin of the gastrocnemius was released at the femoral condyle, leaving a stump of 3–4 cm. The lateral part of the gastrocnemius was also released, and both heads were transferred to the tibial condyles. The released semitendinosus tendon was subsequently sutured to the medial gastrocnemius tendon stump at the medial femoral condyle under slight tension. The rectus femoris was released from its proximal origin at the anterior inferior iliac spine in all CBM patients and sutured to the anterior hip joint capsule, while the reflected head of the rectus femoris muscle was released.

In contrast, in the MTL group the semitendinosus was lengthened by intramuscular tenotomy (middle thigh, supine position) or *Z*-lengthening (popliteal, prone position), and the gastrocnemius muscle was lengthened by intramuscular aponeurotic lengthening. Proximal rectus femoris release was performed only in cases with a double-bump pattern of the pelvis and a positive Duncan-Ely test in the MTL group.

2.4. Postsurgical management

Epidural anaesthesia was used for the first days after surgery, and passive range of motion treatment was started early. All the patients wore lower leg weight-bearing casts for 4 weeks and long-leg night orthoses for 6 months to maintain knee extension. After 4 weeks ankle-foot orthoses with dorsiflexion stop were fitted to assist passive extension of the knee. In cases with additional bony foot surgery (derotation osteotomy or bony foot reconstruction) short-leg non-weight-bearing casts were fitted for 4 weeks after surgery.

2.5. Statistical analysis

Both limbs of each patient were used for further analysis. The GGI [24] was calculated for all subjects at all examinations. Descriptive statistics were used for basic statistical analysis. To show time and group effects, two-way repeated-measures analyses of variance (ANOVA) were applied. p-Values of less than 0.05 were considered to show significant differences, and Bonferroni correction was employed to adjust for multiple comparisons. Statistical analysis was done using PASW® Statistics 18.

2.6. Funding

There was no external source of funding for this investigation.

3. Results

The results of three-dimensional gait analysis and clinical examination are summarized in Table 2 and Figs. 1–3.

3.1. Kinematics

APT increased significantly ($p < 0.01$) in the MTL group directly after surgery, while there was only a moderate, non-significant increase in the CBM group. At E2 both groups showed a decrease: APT values were similar and there was no significant difference between the groups. However, the APT in the MTL group was still

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Table 1
Demographics, global walking ability and surgical procedures.

Parameters	CBM			MTL		
	Baseline (E0)	1 year follow-up (E1)	9 year follow-up (E2)	Baseline (E0)	1 year follow-up (E1)	9 year follow-up (E2)
Gender	8 ♀, 13 ♂	8 ♀, 13 ♂	8 ♀, 13 ♂	6 ♀, 15 ♂	6 ♀, 15 ♂	6 ♀, 15 ♂
Time of follow-up [y]	–	1.3 (±0.6)	9.2 (±2.5)	–	1.2 (±0.7)	9.1 (±2.6)
Age of patients [y]	11.3 (±3.1)	12.6 (±3.2)	20.5 (±4.3)	11.1 (±5.4)	12.3 (±3.7)	20.2 (±4.5)
BMI[a] (kg/m^2)	18.3 (±3.7)	19.4 (±3.6)	20.9 (±4.0)	17.3 (±3.1)	18.1 (±3.1)	21.4 (±4.8)
GMFCS[b]						
GMFCS I	3	6	4	3	7	5
GMFCS II	11	5	9	13	6	10
GMFCS III	7	10	8	5	8	6
GGI[c]	407 (295)$^{\#+}$	227 (160)$^{*+}$	270 (163)$^{*\#}$	364 (175)$^{\#+}$	273 (221)*	233 (121)*

Surgical procedures

Procedures	**CBM**	**MTL**
Intramuscular Psoas lengthening	7	6
Adductorlongusrecession	6	10
Proximal rectus femoris release	42	18
Distal rectus femoris transfer	42	38
Semitendinosus transfer to distal femur	42	0
Intramuscular semitendinosus tenotomy	0	38
Semitendinosus *Z*-lengthening	0	4
Semimembranosus aponeurotic recession	42	42
Lateral hamstring lengthening	6	9
Gastrocnemius transfer to proximal tibia	42	0
Gastroc/soelusaponeurotic recession	12	29
Tendon Achilles lengthening	4	4
Soft tissuefoot	9	9
Femoral derotation osteotomy (proximal)	17	18
Femoral derotation osteotomy (distal)	0	10
Tibialinternalrotationosteotomy	5	5
Bony foot correction	9	15

[a] BMI: Body Mass Index.
[b] GMFCS: Gross Motor Function Classification Scale.
[c] GGI: Gillette Gait Index.
ANOVA (GGI) was used for statistical analysis, Bonferroni corrected; level of significance $p < 0.05$:
*significant difference to pre-operative;
#significant difference to 1 year post-operative;
+significant difference to 9 year post-operative.

greater than at baseline ($p = 0.04$). Minimum hip flexion in stance was improved only in the CBM group, but the decrease was not significant. At E2, the two groups showed identical minimum hip flexion in stance. Range of knee flexion initially increased, but had deteriorated in both groups by E2. Minimum knee flexion in stance, knee flexion at initial contact and peak knee flexion in swing were significantly decreased in both groups, and the decrease was greater in the CBM group. In both groups significant deterioration of both stance phase parameters was found between E1 and E2, and the two groups showed similar values for all kinematic knee parameters at E2, with the exception of peak knee flexion in swing, where a difference was found. The range of dorsi-/plantar flexion was significantly reduced in both groups at E1 and only partial-yet significant-recovery was found in both groups at E2. Peak ankle dorsiflexion in stance was significantly increased in both groups at E1 and remained higher at E2. Cadence was significantly reduced at E1 in both groups, the decrease being larger in the CBM group. However, both groups showed a significant increase in cadence at E2, with higher values in the MTL group. Normalized walking speed was significantly reduced in both groups 1 year after surgery but subsequently recovered, and no difference was found between the two groups at E2. The maximum knee flexor moment was significantly reduced in the CBM group.

3.2. Clinical examination

In both groups hip flexion contracture angle (Thomas test) and popliteal angle were significantly reduced 1 year after surgery but deteriorated significantly between E1 and E2. Passive dorsiflexion had improved in both groups by 1 year after surgery. While the improvement was maintained in the CBM group, a significant deterioration was found at E2 in the MTL group.

3.3. Adverse effects

An APT value more than 2 SD above the norm was classified “increased”. Increased APT was found in 33% of the limbs in the CBM group and 43% in the MTL group at E1. The prevalence of increased APT decreased between E1 and E2 in the MTL group, while it did not change in the CBM group, resulting in almost identical prevalence of increased APT in CBM and MTL patients at long-term follow-up (Fig. 2).

One year after surgery 33% of the limbs (14 of 42: 5 bilateral, 4 unilateral) in the CBM group and 17% (7 of 42: 2 bilateral, 3 unilateral) in the MTL group showed genu recurvatum (knee hyperextension of more than 5° in stance phase). At long-term follow-up genu recurvatum was found in only 5% of the limbs (2 unilateral) in the CBM group and in one limb (2.5%) in the MTL group (Fig. 2).

3.4. Recurrence of flexed knee gait

Recurrence of flexed knee gait, defined as the increase of minimum knee flexion in stance between E1 and E2, was calculated for each limb in both groups. Fig. 3 shows the distribution of recurrence in both groups.

4. Discussion

Since a high level of motor control is needed for normal function of the biarticular muscles of the lower extremity, patients with

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Table 2
Results of three-dimensional gait analysis and clinical examination.

Parameters		E0* (pre)	E1$^{\#}$ (1 year)	E2^{+} (9 years)	Group diff
Instrumented three-dimensional gait analysis					
Gillette Gait Index	CBM	407 (295)$^{\#+}$	227 (160)$^{*+}$	270 (163)$^{*\#}$	–
	MTL	364 (175)$^{\#+}$	273 (221)*	233 (121)*	
Mean pelvic tilt	CBM	15 (6)	19 (7)$^{+}$	17 (8)$^{\#}$	–
	MTL	14 (8)$^{\#+}$	21 (8)$^{*+}$	17 (7)$^{*\#}$	
Minimum hip flexion in stance	CBM	10 (15)	4 (10)	6 (11)	–
	MTL	6 (11)	6 (14)	7 (11)	
Range of knee flexion/extension	CBM	30 (13)$^{\#+}$	44 (11)$^{*+}$	39 (12)$^{*\#}$	–
	MTL	33 (13)$^{\#+}$	48 (12)$^{*+}$	40 (11)$^{*\#}$	
Knee flexion at initial contact	CBM	41 (14)$^{\#+}$	16 (10)$^{*+}$	24 (8)$^{*\#}$	E1
	MTL	41 (16)$^{\#+}$	23 (6)$^{*+}$	27 (8)$^{*\#}$	
Minimum knee flexion in stance	CBM	28 (20)$^{\#+}$	5 (14)$^{*+}$	13 (11)$^{*\#}$	–
	MTL	28 (21)$^{\#+}$	7 (13)$^{*+}$	16 (10)$^{*\#}$	
Peak knee flexion in swing	CBM	58 (12)$^{\#+}$	49 (10)*	52 (9)*	E1, E2
	MTL	61 (13)$^{\#+}$	56 (8)*	56 (7)*	
Range of dorsi/plantar flexion	CBM	29 (13)$^{\#+}$	20 (7)$^{*+}$	21 (7)$^{*\#}$	–
	MTL	25 (8)$^{\#+}$	18 (5)$^{*+}$	23 (7)$^{*\#}$	
Peak dorsiflexion in stance	CBM	9 (18)$^{\#+}$	14 (8)*	15 (6)*	E2
	MTL	8 (13)$^{\#+}$	12 (6)*	12 (7)*	
Normalized cadence[a]	CBM	43.3 (8.7)$^{\#}$	35.5 (10.6)$^{*+}$	41.8 (9.7)$^{\#}$	E2
	MTL	42.9 (5.6)$^{\#}$	37.9 (10.8)$^{*+}$	45.6 (6.2)$^{\#}$	
Normalized speed[a]	CBM	0.22 (0.07)$^{\#}$	0.19 (0.08)$^{*+}$	0.20 (0.08)$^{\#}$	–
	MTL	0.22 (0.06)$^{\#}$	0.19 (0.09)$^{*+}$	0.22 (0.05)$^{\#}$	
Normalized step length[a]	CBM	0.30 (0.07)	0.30 (0.07)	0.28 (0.05)	–
	MTL	0.31 (0.05)	0.29 (0.08)	0.28 (0.05)	
Minimum knee flexor moment	CBM (n = 16)	−0.38 (0.83)	−0.48 (0.25)	−0.53 (0.26)	–
	MTL (n = 20)	−0.36 (0.16)	−0.41 (0.11)	−0.42 (0.11)	
Maximum knee flexor moment	CBM (n = 16)	0.73 (0.35)$^{\#}$	0.41 (0.23)*	0.52 (0.31)	–
	MTL (n = 20)	0.70 (0.38)	0.48 (0.21)	0.58 (0.31)	
Maximum DorsiPlantFlex moment	CBM (n = 16)	1.22 (0.35)	1.17 (0.26)	1.25 (0.17)	E0, E1
	MTL (n = 20)	1.00 (0.35)	1.02 (0.22)$^{+}$	1.14 (0.17)$^{\#}$	
Clinical examination					
Thomas test	CBM	15 (10)$^{\#+}$	5 (7)$^{*+}$	9 (7)$^{*\#}$	–
	MTL	12 (13)$^{\#+}$	5 (6)$^{*+}$	6 (7)$^{*\#}$	
Popliteal angle	CBM	48 (17)$^{\#}$	22 (14)$^{*+}$	54 (16) $^{\#}$	–
	MTL	51 (21)$^{\#}$	32 (18)$^{*+}$	53 (15)$^{\#}$	
Passive knee extension	CBM	−7 (6)$^{\#+}$	0 (2)*	−2 (8)*	–
	MTL	−9 (13)$^{\#+}$	−1 (6)*	−1 (6)*	
Passive ankle dorsiflexion	CBM	−1 (13)$^{\#}$	7 (8)$^{*+}$	5 (7)$^{\#}$	E2
	MTL	−0.4 (14)$^{\#}$	4 (7)$^{*+}$	0.4 (8)$^{\#}$	
Passive ankle plantar flexion	CBM	38 (5)$^{\#}$	39 (6)$^{*+}$	34 (10)*	E1, E2
	MTL	43 (11)$^{\#}$	35 (11)*	35 (18)$^{\#}$	

Mean values ± (SD) preoperatively and 1 and 9 years postoperatively for all limbs of the CBM (conversion of biarticular muscles) and the MTL (muscle-tendon lengthening) group are shown for gait-analysis parameters; two-way ANOVA (factor GROUP and factor TIME) was used for statistical analysis, Bonferroni corrected; level of significance $p < 0.05$:
*significant difference from preoperative;
$^{\#}$significant difference from 1 year postoperative;
$^{+}$significant difference from 9 year postoperative.
[a] Spatio-temporal parameters step length, cadence and walking speed rendered dimensionless using the normalization variable "stature" [m]according to Hof et al. [25].

cerebral palsy tend to have more difficulty controlling biarticular than monoarticular muscles [15,16,26]. The disturbed function of the biarticular muscles in cerebral palsy is of major importance in the development of gait abnormalities such as flexed knee gait. These abnormalities in the function of biarticular muscles can be modified by MTL as a component of SEMLS, and various studies have reported satisfactory results after hamstring lengthening [6,8–13,27]. However, increased APT due to hamstring weakness and recurrence of flexed knee gait have been reported as adverse effects [9,10,13,27]. This may partially be explained by the fact that some patients with flexed knee gait have normal or even increased hamstring length [28]. Hamstring lengthening in these patients may lead to muscle weakness and APT. Increased APT has been suggested to be a major factor leading to recurrence of flexed knee gait [27]. CBM aims to reduce these adverse effects, but its surgical impact is considerably higher than that of MTL.

In our study CBM resulted in a better overall kinematic outcome than MTL 1 year after surgery, reflected in less APT, greater improvement of knee extension in stance and a smaller popliteal angle. Despite concomitant lengthening of semimembranosus and in a few cases biceps femoris there was no significant increase in APT in the CBM group, while the patients of the MTL group showed a significant increase in mean APT. It seems that the transfer of the semitendinosus tendon has the potential to stabilize the pelvis. The short-term efficacy of CBM is in accordance with previous reports [16,22]. However, most of the patients in past studies were not followed up after their main growth spurt and changes beyond the scope of these studies may influence the outcome, since deterioration of gait parameters during growth is possible in children with cerebral palsy [29]. It is therefore of major interest whether these advantages of CBM are maintained until the end of growth.

These favourable effects of CBM in our study vanished at long-term follow-up, resulting in comparable overall kinematic and clinical function in the two groups. This shows that CBM, a significantly more extensive intervention for the patient and surgeon, has no long-term advantage over MTL. When considering each individual patient, the prevalence of increased APT was higher in the MTL group than in the CBM group one year after surgery. This may be explained by the weakening and over-lengthening of the hamstrings in the MTL group, while it seems

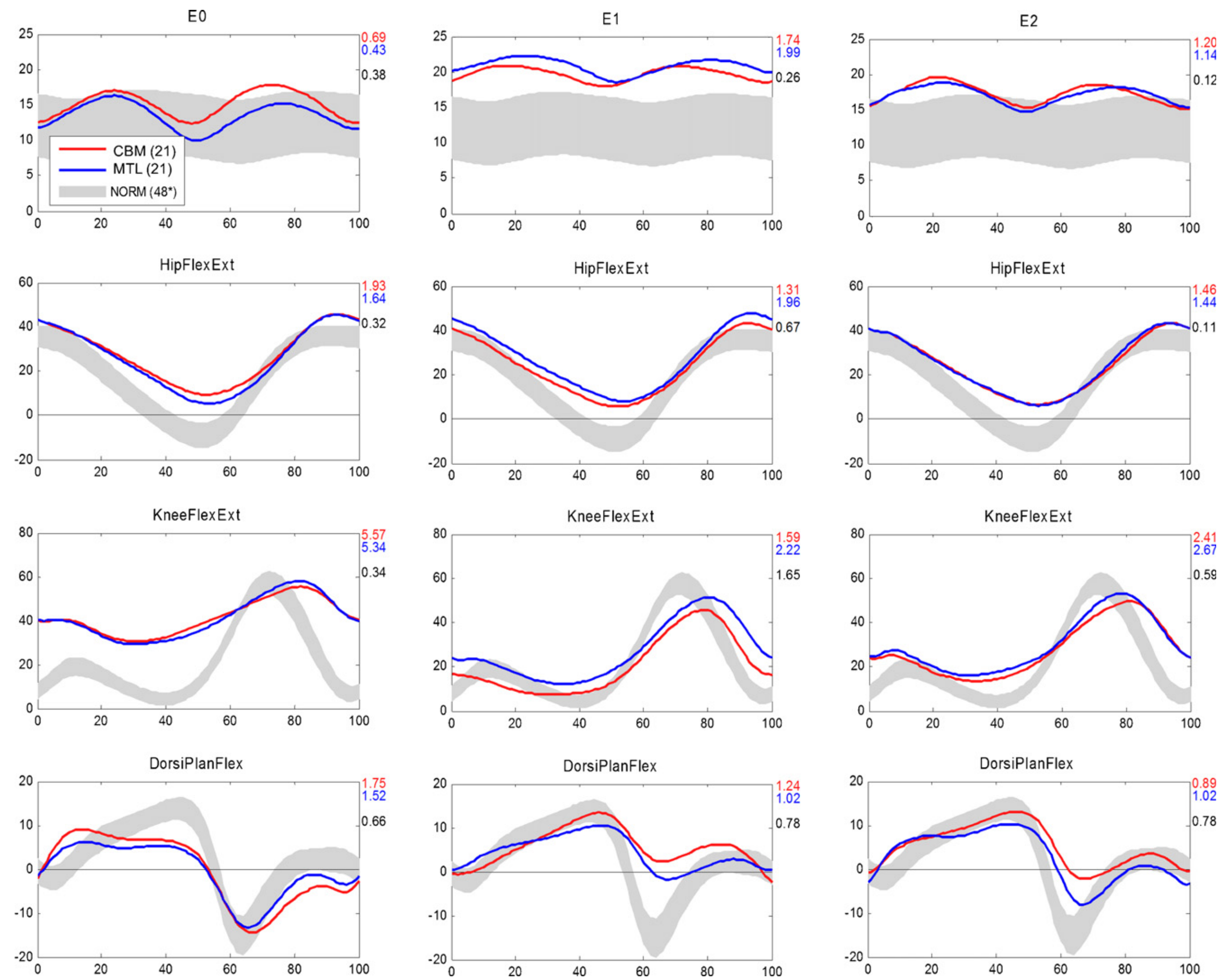

Fig. 1. Sagittal plane knee kinematics. Average sagittal plane kinematic mass graphs of pelvic tilt (1st line), hip flexion (2nd line), knee flexion (3rd line) and ankle dorsi/plantar flexion (4th line) for all patients of the CBM group (red line) and the MTL group (blue line) as well as for an age-matched group of 48 norm subjects, represented by the grey area (including 1 standard deviation), are shown. Graphs are visualized for all examinations: preoperative (E0, 1st column), 1 year post- (E1, 2nd column) and 9 years postoperative (E2, 3rd column). Positive values indicate anterior pelvic tilt, hip flexion, knee flexion and ankle dorsiflexion. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

that the transfer of the semitendinosus tendon to the femur has some stabilizing effect on the pelvis in the CBM group. However, the prevalence of increased APT in both groups is excessively high and cannot be left unremarked. Both procedures are accompanied by significant weakening of the hamstrings, leading to loss of pelvic stability in the sagittal plane. This increase in APT and the conversion of the gastrocnemius muscle may explain why the CBM group tended to display genu recurvatum 1 year after surgery more often than the MTL group. Genu recurvatum was significantly less frequent at long-term follow-up, when both groups showed very low prevalence of knee hyperextension. However, the prevalence of increased APT did not change between E1 and E2 in the CBM group and only a marginal reduction was found in MTL group. These long-term results indicate that a substantial increase in mean APT has to be expected after any surgery, only partially recovering over time. Since aponeurotic semimembranosus lengthening was done in all patients of both groups and lateral hamstring lengthening was needed in a few cases, persistent

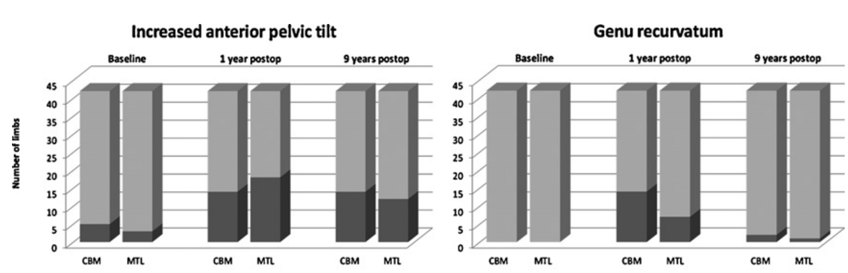

Fig. 2. Adverse effects. The prevalence of increased anterior pelvic tilt (APT) and genu recurvatum among all limbs is shown for the CBM and the MTL group at the 1 year (E1) and the 9 year (E2) examination. Dark grey bars represent the affected limbs while light grey bars represent all other limbs.

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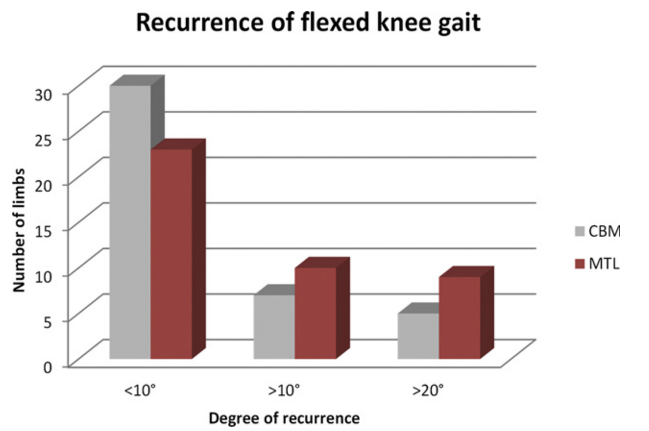

Fig. 3. Recurrence of flexed knee gait. The amount of recurrence of flexed knee gait was calculated by the difference between minimum knee flexion in stance at E1 and E2 for each individual leg for all patients of both groups. According to this difference, the legs were categorized into three groups (abscissa): recurrence less than 10° (1st bar), recurrence of more than 10° (2nd bar), recurrence of more than 20° (3rd bar). The number of legs is shown on the axis of ordinates.

hamstring over-length and weakness has to be taken into consideration as the major cause for increased APT. Persistent increased APT should be considered as one possible factor leading to partial recurrence of flexed knee gait as found in this study [27]. However, since the overall APT improved in the MTL group, some patients must have the potential to recover muscle strength and sufficiently stabilize the pelvis despite treatment by MTL, or muscle length is re-established as an effect of growth. It may therefore be meaningful to ask which patients are prone to develop increased APT. In accordance with the suggestion of Ma et al. [22] there is a general belief that patients with a higher GMFCS level, who are limited concerning compensation of muscle weakness, are particularly prone to adverse effects and recurrence of flexed knee gait. The patients in the present study were also matched according to GMFCS level in order to compare patients with the same degree of severity. We found no correlation between GMFCS level and development of increased APT. However, this may be due to the relatively small sample sizes. With regard to recurrence of flexed knee gait, both groups showed a significant deterioration between the 1-year and long-term examinations, while the MTL group exhibited a non-significant tendency towards greater deterioration. The long-term recurrence of flexed knee gait seems to be influenced by increased APT. Since CBM did not yield superior results and its surgical impact on the patient is considerably higher than that of MTL, CBM cannot be recommended.

One recently described treatment concept for flexed knee gait correction comprises femoral extension osteotomy and patella tendon shortening [30]. In the absence of long-term results, however, this procedure cannot yet be compared with MTL and CBM.

5. Limitations

During SEMLS different surgical procedures are commonly performed in one session. Therefore, the concomitant procedures may have affected the results in both patient groups in our study. Although we strove to minimize these influences by matching the patients according to different gait analysis parameters, this factor should be considered when interpreting the results.

Conflict of interest

None.

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The effects of muscle-tendon surgery on dynamic electromyographic patterns and muscle tone in children with cerebral palsy

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ABSTRACT

During multilevel surgery, muscle-tendon lengthening (MTL) is commonly carried out in children with cerebral palsy. However, it is unclear if MTL also modifies increased muscle tone and if pathologic activation patterns are changed as an indirect effect of the biomechanical changes. Since investigations addressing this issue are limited, this study aimed at evaluating the effects of MTL on muscle tone and activation pattern. Forty-two children with spastic diplegia who were treated by MTL underwent standardized muscle tone testing (modified Ashworth and Tardieu test), dynamic EMG and three-dimensional gait analysis before, one and three years after MTL. For the evaluation of muscle activation patterns the norm-distance of dynamic EMG data was analyzed. Range of motion and joint alignment in clinical examination were found to be significantly improved one year after MTL. However, deterioration of these parameters was noted after three years. Muscle tone was significantly reduced one year postoperatively but showed an increase after three years. Joint kinematics were found significantly closer to reference data of age matched controls initially after surgery, but deteriorated until three years postoperatively. However, the EMG patterns of the muscles which were surgically addressed were found to be unchanged in either follow-up. These findings suggest that despite the influence of MTS on biomechanics and physiology (muscle tone reduction and improvements of joint mobility and gait pattern) MTS does not change abnormal patterns of muscle activation. Recurrence of increased muscle tone and deterioration of kinematic parameters three years after surgery may be attributed to these persistent pathologic activation patterns.

1. Introduction

Apart from secondary causes such as lever-arm dysfunction, abnormal muscle tone is accused to be a primary factor for gait disorders in the presence of cerebral palsy [1–4]. A common approach for the treatment of dynamically and structurally shortened muscles is muscle-tendon lengthening (MTL), of which effectiveness was shown [1–5]. Another effect expected after MTL is a reduction of abnormal muscle tone. Increased muscle tone summarizes two effects induced by impaired central regulation [1]: the enduring muscle rigidity due to permanent overstimulation of muscle fibers and the sudden increase in muscle resistance due to hyper-excitability of muscle-tendon reflexes as a velocity dependent finding which is a primary feature associated with spasticity [6]. The first effect is primarily tested by the modified Ashworth [7] test the latter is assessed by the Tardieu [8] test. If there is a velocity dependent hypertonic reflex in spastic muscles as described by Lance [6], MTL theoretically may have the potential to decrease tone by de-tensioning the spindle receptors in tendons and muscles. Further, altered biomechanics may lead to altered stabilization and in consequence altered muscle (co-) activation.

In this context, Granata et al. could find significant changes in EMG patterns of the gastrocnemius–soleus complex, while proximal patterns remained unchanged [9]. In contrast, only marginal changes were found by Patikas et al. [10].

Furthermore, there is no knowledge how muscle tone and activation develops beyond a short-term period following MTL, especially when considering possible recurrence of increased muscle tone and gait disorders in CP children [11–13]. Therefore, the hypothesis of this study was that MTL reduces tone of key muscles as assessed by Tardieu and Ashworth test and that the surgery increases joint ranges of motion. Hence it may modify the pathologic patterns of muscle activation in children with spastic diplegia monitored in dynamic EMG. Furthermore, the study intended to evaluate the development of muscle tone and

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activation patterns on a midterm basis up to 3 years after MTL, when possible recurrences of biomechanical disorders with influence on gait may be expected.

2. Methods

This study included children with spastic diplegic CP selected from the gait lab database treated by single-event multilevel surgery including MTL of biarticular muscles (gastrocnemius and hamstrings). Inclusion criteria were a standardized clinical examination including modified Ashworth [7] and Tardieu test [8] muscle tone testing and 3D-GA pre-, one and three years post MTL surgery. Exclusion criteria were previous surgery, Botulinum-toxin-injection less than six months prior to MTL or between the examinations, dystonic CP and severe mental retardation.

Forty-two children were included. The age at the time of MTL and pre-operative examination ranged from 6 to 16 years (9.8 ± 2.8 years). For details of demographic data see Table 1. All the patients were treated by MTL (Table 1). 3D-GA including kinematics and dynamic EMG as well as clinical examination (passive range of motion and muscle tone testing according to the modified Ashworth [7] and Tardieu [8] test) were carried out preoperatively (E0), one year (E1, 0.9 ± 0.2) and three years (E2, 3.2 ± 1.1) after surgery. The degree of increased muscle tone was measured according to previous reports [7,8].

- modified Ashworth test:
- 0: normal tone
- 1: slight increase in tone, a catch and release at the end of range of motion
- 1+: slight increase in tone, a catch followed by minimal resistance in remainder of range
- 2: more marked increase in muscle tone through most of the ROM, but affected part(s) easily moved

Table 1
Demographics and surgical procedures.

Parameters	E0 (pre-op)	E1 (1 y post)	E2 (2–4 y post)
Sex: ♀ = 15 ♂ = 27			
Time of follow-up (years)		0.9 (± 0.2)	3.2 (± 1.2)
Age of patients (years)	9.8 (± 2.8)	10.9 (± 2.8)	13.1 (± 2.5)
BMI	17.3 (± 3.0)	18.0 (± 3.5)	19.3 (± 3.7)
GMFCS I	6	8	10
GMFCS II	28	23	22
GMFCS III	8	11	10

Procedures	All legs (n = 84 legs)	
	Left side	Right side
Psoas over the brim	8	6
Proximal rectus femoris release	10	12
Medial hamstring lengthening	42	42
Lateral hamstring lengthening	12	10
Distal rectus femoris transfer	40	38
Gastrocnemius/soleus intramuscular recession	42	40
Femoral derotation osteotomy	31	32
Tibia derotation osteotomy	5	6
Bony foot stabilization (Evans, Grice, Chopart fusion)	17	14

GMFCS (gross motor function classification scale).

Table 2
Mean (SD) values (in degrees) and statistical results for specific parameters.

Parameters	E0 (pre-op)	E1 (1 y post)	E2 (3 y post)
Clinical examination			
Thomas test1	10 (40)[b,c]	0 (15)[a,c]	5 (35)[a,b]
Passive hip extension1	3 (40)	5 (25)	10 (30)
Passive hip flexion1	120 (55)[b,c]	117.5 (50)[a]	110 (110)[a]
Popliteal angle1	52 (18)	38 (16)[a,c]	45 (18)[a,b]
Passive knee extension1	−6 (11)[b,c]	−0.1 (6)[a]	−0.4 (6)[a]
Passive knee flexion1	150 (9)[b,c]	146 (10)[a]	142 (12)[a]
Passive dorsiflexion (knee in 90° flexion)1	7 (13)[b,c]	14 (8)[a]	13 (10)[a]
Passive dorsiflexion (knee in extension)1	44 (9)[b,c]	37 (10)[a]	39 (9)[a]
Passive plantarflexion (knee in extension)1	44 (11)[b,c]	35 (9)[a]	36 (10)[a]
Tardieu- test2	1 (3)[b,c]	0 (3)[a]	0 (3)[a]
Hamstring tone2	1 (0.8)[b,c]	0.5 (0.6)[a]	0.8 (0.6)[a,b]
Triceps surae tone (knee in 90° flexion)2	1.5 (3)[b,c]	1.0 (2)[a]	1.0 (2)[a]
Triceps surae tone (knee in extension)2	1.5 (3)[b,c]	1.0 (2)[a]	1.25 (3)[a]
Three-dimensional gait analysis			
Minimum hip flexion in stance1	4 (11)	4 (11)	3 (10)
Mean hip flexion in stance1	21 (9)	19 (9)	19 (9)
Knee flexion at initial contact1	35 (13)[b,c]	19 (10)[a]	21 (7)[a]
Minimum knee flexion in stance1	17 (19)[b,c]	3 (12)[a]	6 (12)[a]
Mean knee flexion in stance1	27 (17)[b,c]	13 (11)[a]	16 (11)[a]
Range of knee flexion1	39 (15)[b,c]	51 (12)[a]	49 (13)[a]
Peak knee flexion in swing1	56 (13)	54 (9)	55 (9)
Dorsiflexion at initial contact1	−8 (12)[b,c]	−2 (5)[a]	−4 (6)[a]
Peak dorsiflexion in midstance1	2 (15)	7 (6)	7 (7)
Mean dorsiflexion in midstance1	−1 (15)c	5 (6)	5 (7)[a]
Minimum dorsiflexion in stance1	−25 (18)[b,c]	−8 (6)[a,c]	−11 (8)[a,b]
Peak dorsiflexion in swing1	−7 (13)[b,c]	3 (5)[a]	2 (7)[a]
Dynamic EMG			
Vastus lateralis1	1.1 (0.3)	1.2 (0.4)	1.2 (0.3)
Rectus femoris1	1.2 (0.2)	1.2 (0.3)	1.2 (0.3)
Semimembranosus1	0.8 (0.2)	0.8 (0.2)	0.8 (0.2)
Biceps femoris1	0.9 (0.2)	0.9 (0.2)	0.9 (0.2)
Gastrocnemius1	1.3 (0.3)	1.2 (0.3)	1.2 (0.2)
Soleus1	1.1 (0.3)[b,c]	0.9 (0.3)[a]	0.8 (0.2)[a]
Tibialis anterior1	1.3 (0.3)	1.3 (0.4)	1.2 (0.4)

1 = Anova and Bonferroni correction (mean and standard deviation). 2 = Wilcoxon test (median and range).
[a] Significant difference from E0.
[b] Significant difference from E1.
[c] Significant difference from E2.

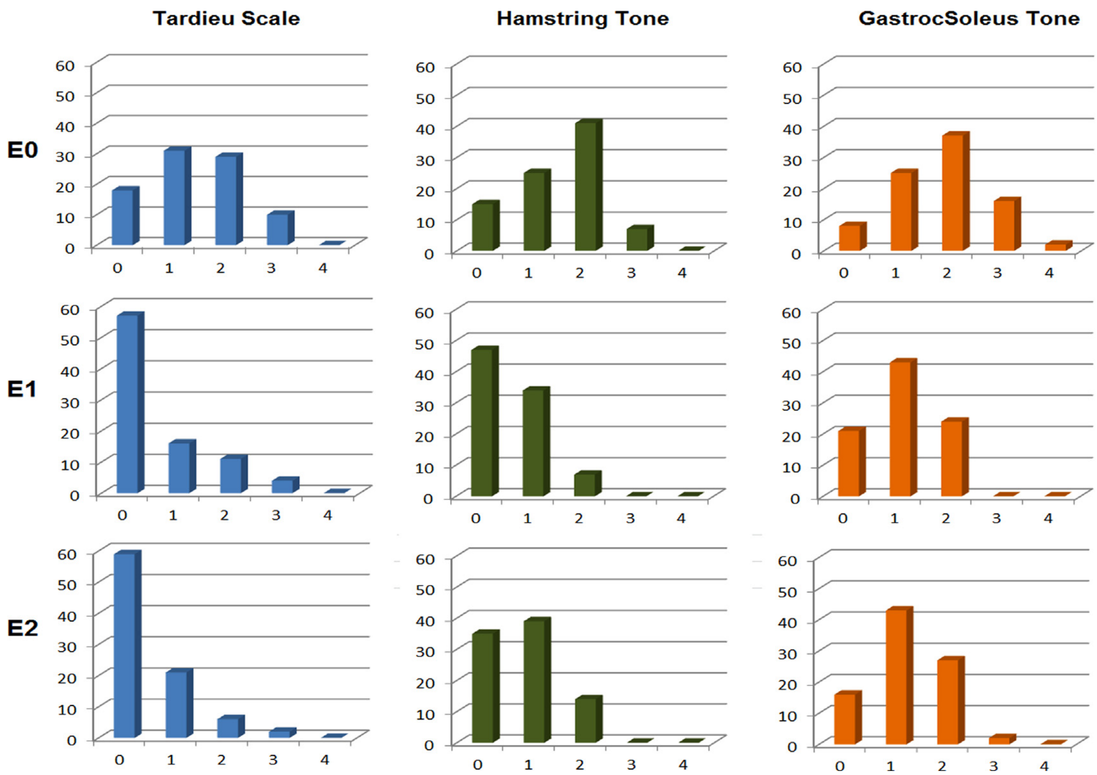

Fig. 1. Development of muscle tone. Distribution of Tardieu scale (1st column), Ashworth scale for hamstrings (2nd column) and Ashworth scale for gastroc-soleus-complex (3rd column, measured in knee extension) are displayed at before (1st line, E0) as well as one year (2nd line, E1) and three years (3rd line, E2) after surgical intervention. Modified Ashworth scale: 0: normal tone; 1: slight increase in tone, a catch and release at the end of range of motion; 1+: slight increase in tone, a catch followed by minimal resistance in remainder of range; 2: more marked increase in muscle tone through most of the ROM, but affected part(s) easily moved; 3: considerable increase in tone, passive movement difficult; 4: affected parts rigid in flexion or extension. Ashworth level 1 and 1+ are shown summarized as "1".

- 3: considerable increase in tone, passive movement difficult
- 4: affected parts rigid in flexion or extension.

\- modified Tardieu test:

- 0: no resistance throughout the course of passive motion
- 1: mild resistance slowing down the passive movement, no clear catch at a precise angle
- 2: clear catch during course of passive motion at a precise angle followed by a release
- 3: fatigable clonus with less than 10 s when maintaining the pressure appearing at a precise angle
- 4: unfatigable clonus with more than 10 s when maintaining the pressure and appearing at a precise angle.

For standardized 3D-GA, the patients walked barefoot along a line 7 m in length during data acquisition. A 120 Hz nine camera system (Vicon®, Oxford Metrics, UK) and reflecting markers placed at well defined anatomical landmarks were used according to the standardized protocol of Kabada et al. [14].

For dynamic EMG, data from the following key muscles were collected: rectus femoris, vastus lateralis, semimembranosus, biceps femoris, gastrocnemius and soleus muscles. Each muscle was applied with bipolar surfaces adhesive electrodes (Blue Sensor, Ambu Inc., Glen Burnie, MD, USA) according to the SENIAM guidelines [15]. Pre-amplification of the resulting EMG signal was done by using the Biovision EMG apparatus (Biovision Inc., Wehrheim, Germany). EMG data were digitized according to previous reports [10] using a 16-bit A/D card with the sample frequency of 1080 Hz. The EMG signals were fully commutated off-line and the linear envelopes were calculated at the cut-off frequency of 9 Hz [16]. The EMG amplitudes were normalized to the mean value for each muscle of each step and subject respectively. Time normalization of EMG data was performed for stance and swing phase independently to account for different gait cycle timing between subjects with CP and the reference group of 20 typically developing age-matched children.

2.1. Data analysis and statistical analysis

For assessing the similarity of the individual EMG pattern to the average EMG pattern of the age-matched reference group, the distance between normal and pathologic data was calculated according to a previous report [10]. This measure is defined as the absolute difference between the EMG pattern of the individual patient (p) and the mean value of the norm group (N) at a specific time (t), normalized to the standard deviation (s) of the norm group at the same time (t).

$$\mathrm{ND}_{pt} = \frac{|x_{pt} - x_{Nt}|}{s_{Nt}}$$

In order to evaluate the deviation from normal of each parameter, we quantified our results by calculating the mean norm-distance for the whole gait cycle.

Only left limbs were used for statistical analysis. For comparisons of the repeated measures data, one-way repeated measures analyses of variance (ANOVA) were applied to show significant differences between the examinations. For ordinal data, Friedmann test with post hoc Wilcoxon signed-rank tests was conducted. P-values of less than 0.05 were treated to be significant. Bonferroni correction was used to adjust for multiple comparisons. Unless otherwise stated, statistical analysis of the results was done using PASW Statistics 18.

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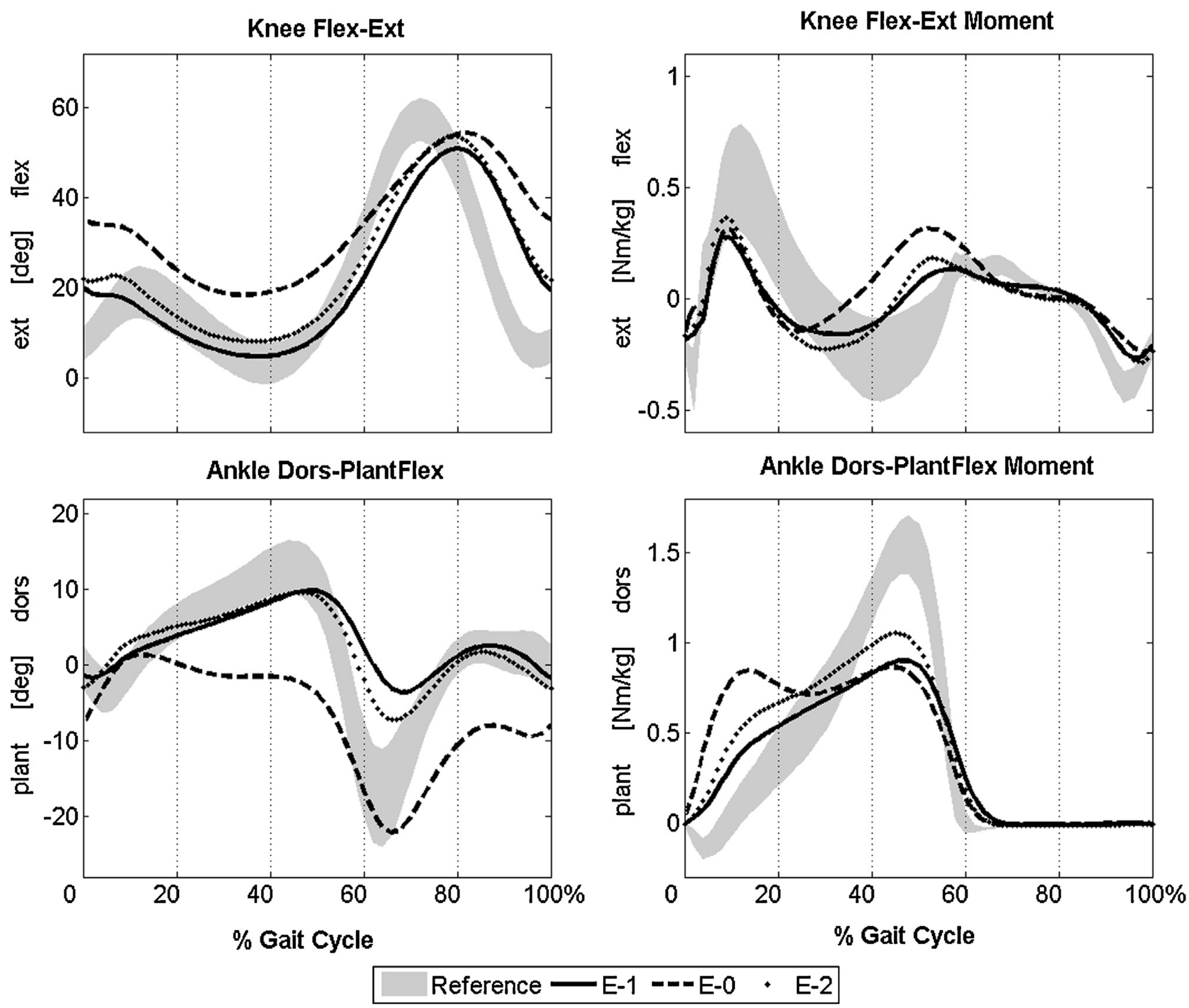

Fig. 2. Sagittal knee and ankle kinematics and kinetics. Average sagittal plane kinematic (1st column) and kinetic (2nd column) mass graphs of the knee (1st line) and ankle (2nd line) for all the patients are shown. Graphs are visualized for all examinations: preoperative (E0, dashed line), 1 year post (E1, solid line) and 3 years (E2, dotted line) post intervention. In all graphs the reference data including obtained by an age-matched group of 25 norm subjects is represented by the grey area (including 1 standard deviation). Positive values indicate knee flexion/ankle dorsiflexion.

3. Results

The development of specific parameters from clinical examination, 3D-GA and dynamic EMG are summarized in Table 2.

3.1. Clinical exam

3.1.1. Passive range of motion

A significant reduction of popliteal angle could be found at E1. Between E1 and E2 popliteal angle showed a significant deterioration. Passive knee extension was found to be increased significantly at E1 and this was maintained at E2, while passive knee flexion was slightly reduced. A significant improvement of passive dorsiflexion was found after surgery and this improvement was maintained at E2. This was accompanied by a permanent reduction of passive plantarflexion.

3.1.2. Muscle tone assessment

A significant reduction in muscle tone in respect of the modified Ashworth and Tardieu testing of the hamstrings was found at E1 (Fig. 1). At E2 there was a significant increase of hamstring muscle tone (Fig. 1). The triceps surae tone was found to be significantly reduced at E1 but tended to increase between E1 and E2 (Fig. 1).

3.2. Three-dimensional gait analysis

3.2.1. Kinematics

Knee flexion at initial contact, mean and minimum knee flexion in stance were significantly reduced at E1. In all three parameters a tendency for deterioration was found at E2 which was significant for minimum and mean knee flexion in stance. The total range of knee motion was found to be significantly improved at E1 and showed a slight deterioration between E1 and E2. Peak knee flexion in swing did not change significantly (Fig. 2).

Ankle dorsiflexion was significantly improved at initial contact, during midstance and during swing phase. This improvement was maintained at E2. This was accompanied by a significant reduction of plantar flexion and total ROM (Figure II).

3.2.2. Dynamic Electromyography

Fig. 2 summarizes the EMG data averaged over the patient group, which was normalized to gait cycle (Fig. 3). A large

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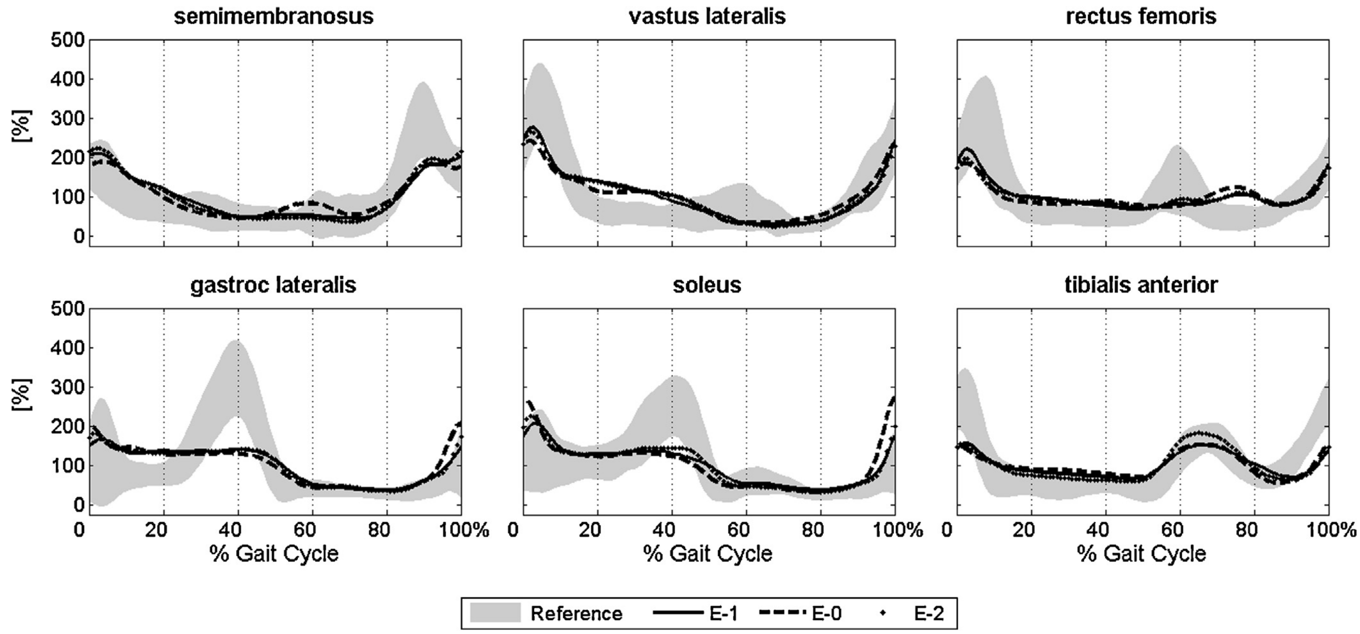

Fig. 3. Dynamic electromyography. Mean EMG data of typical muscles of thigh (semimembranosus, vastus lateralis and rectus femoris) and the lower leg (gastrocnemius/soleus-complex) for all patients are shown. The EMG amplitudes were normalized to the mean value of each muscle of each step and subject respectively. Graphs are visualized for all examinations: preoperative (E0, dashed line), 1 year post- (E1, solid line) and 3 years (E2, dotted line) post intervention. In all graphs the reference data including obtained by an age-matched group of 25 norm subjects is represented by the grey area (including 1 standard deviation).

norm-distance was found pre-operatively for all muscles. The norm-distance of EMG data of all muscles with exception of soleus muscles was not found to be changed significantly at E1. There were also no significant changes found between E1 and E2. For soleus muscle, a marginal but significant reduction of norm-distance (0.1) was found at E1 and there was another but not significant reduction (0.1) between E1 and E2 (Fig. 3).

4. Discussion

Primary reason for MTL is the change of unit length which allows for a more normal gait at mid- [17,18] and long-term [12,13]. This may in secondary consequence normalize EMG patterns. Significant improvements in specific parameters of the clinical examination and 3D-GA were found in our study one year after surgery corroborating previous results [1–4,10,11,17–19] Another result of MTL is a decrease of pretension which reduces force at a given length and thus resistance against stretch during clinical examination visible both in Tardieu and Ashworth testing with possible primary consequences for dynamic EMG patterns.

Clinical assessment of increased muscle tone grades the resistance of the muscle felt by stretching a joint. The factors which obviously determine resistance are muscle stiffness (passive) and muscle tone (active). The latter depend on the evocation of the reflex but also on the force of the responding muscle contraction. By MTL the structural shortness is corrected and thereby the passive part – muscle stiffness – is less relevant. Further, the muscle fibers are de-tensioned which reduces the force of the active contraction (at least at the same angle as before surgery). The same is true for the muscle spindles which may contribute to the active muscle contraction as a reaction on stretch. Indeed, MTL does reduce increased muscle tone significantly affecting both passive and active dynamic components of muscle tightness in children with cerebral palsy one year after surgery [19]. This is underlined by the findings in our study.

In contrast, the reduction of muscle tone does not seem to match a change in muscle activation pattern seen in dynamic EMG. Patikas et al. classified and evaluated the patterns of dynamic electromyography in children with spastic cerebral palsy following multilevel surgery [10]. They could find only minimal changes (EMG patterns of the soleus, gastrocnemius and tibialis anterior muscles got closer to normal) [10]. In earlier studies changes in EMG patterns after surgical intervention were reported but found not to be significant [10,20–22]. In this study, we could not find any significant changes of norm-distance in EMG patterns of vastus lateralis, hamstrings, rectus femoris and gastrocnemius. Hence also timing and dynamics (peak-to-peak) did not change. Only soleus muscle showed a significant but marginal improvement. This is contrary to what Buurke et al. found after hamstring lengthening (delayed off-time of the semitendinosus and decreased burst duration of vastus lateralis) [23]. Granata et al. reported comparable results to ours concerning hip and knee muscles, but found significant alterations in gastrocnemius–soleus complex [9] The results of our study, however, showed that the biarticular gastrocnemius did not change in comparison to the monoarticular soleus muscle when EMG is done separately for both muscles.

The EMG signal must be seen as a compound of the necessary muscle activation for the respective posture, the reflex activity and the central pattern. Hence, the EMG signal only very roughly correlates with muscle strength. It may well be that the effect of weakening due to MTL may lead to a reduction felt by active resistance without changing the EMG. It may further be that the biomechanical situation is not changed enough to change the activation pattern, which overrides the effect of MTL on spastic reflex activity.

To our knowledge there are no studies investigating the further postsurgical development of muscle tone and EMG activation patterns beyond two years. We could find a general increase of muscle tone measured by Tardieu and Ashworth in our collective between E1 and E2. This was accompanied by a deterioration of clinical and 3D-GA parameters indicating that increased tone may be responsible for recurrence. However, Tedroff et al. found that a valid permanent reduction of increased muscle tone caused by

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selective dorsal rhizotomy (SDR) did not prevent contractures in 10 year follow-up [24]. Strange enough, changes in dynamic EMG are rarely found even after SDR [25] and we still lack an explanation. The missing relevant effects for calf muscles may be explained by sparing the rootles of these muscles during SDR in order to avoid bladder problems. However, the effects for thigh muscles remained also unchanged. The questions arise if these unchanged pathologic patterns may have an influence on the development of muscle tone and biomechanical alignment.

There are manifest limitations for both the modified Ashworth and the Tardieu test in the assessment of muscle tone [8,26]. Ideally, the Ashworth test would be sensitive and selective for the enduring muscle rigidity due to permanent overstimulation of muscle fibers as opposed to the Tardieu test which would be sensitive and selective for the sudden increase in muscle resistance due to hyper-excitability of muscle-tendon reflexes as a velocity dependent aspect of spasticity. Differences in outcome between both measures in the course of E1 and E2 of this study could then have possibly elucidated the specific mechanisms by which MTL influences the pathogenesis of cerebral palsy. However, it rather appears that in the simple clinical setting both tests are little selective – both decrease at E1 and increase at E2 – and possibly reflect mainly the alterations in muscle structure rather than muscle tone.

Instrumented testing in an objective manner as done e.g. in the work of van den Noort et al. where they monitored the catch due to spasticity in CP on the basis of EMG monitoring in a clinical setting [27] may improve the selectivity. Such methods [27,28] represent then a relevant approach for further studies investigating the underlying mechanisms effecting muscle tone by muscle tendon surgery.

5. Conclusions

The findings of our study underline that MTS leads to significant reduction of muscle tone (measured by modified Ashworth and Tardieu test) accompanied by significant improvements of joint mobility and biomechanical improvements during gait. However, MTS does not modulate abnormal muscle activation patterns in dynamic EMG. Tone reduction does not coincide with changes in dynamic EMG. Recurrence of increased muscle tone and deterioration of 3D-GA parameters three years after surgery may be attributed to these persistent pathologic patterns of muscle activation.

Conflict of interest statement

We disclose any financial and personal relationships with other people or organisations that could inappropriately influence (bias) this work.

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