PRINCIPLES OF PATHOLOGICAL GAIT

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Katrin Irina Schweizer

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Genehmigt von der Medizinischen Fakultät auf Antrag von:

Prof. Dr. Bert Müller, Fakultätsverantwortlicher & Korreferent

Prof. Dr. med. Reinald Brunner, Dissertationsleiter

Prof. Dr. Urs Granacher, Externer Gutachter

Prof. Dr. Arno Schmidt-Trucksäss, Prüfungsvorsitzernder

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Prof. Dr. Christoph Beglinger
Dekan

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GERMAN SUMMARY

Das bipedale Gehen ist für gesunde Menschen eine einfache und alltägliche Bewegung. Das Gangmuster von Patienten kann jedoch stark von einem gesunden Gang abweichen, vor allem bei Patienten mit neuro-muskulären Erkrankungen. Für den Kliniker ist es essentiell, zu unterscheiden, ob eine Gangbildabweichung primär oder sekundär ist, um mit Hilfe der Ganganalysedaten eine geeignete Behandlung einzuleiten. Primäre Gangbildabweichungen werden als ursächlich angesehen und erfordern eine Behandlung. Sekundäre Abweichungen hingegen bedürfen, unabhängig davon, ob ihnen eine aktive Kompensation oder ein passiver physikalischer Effekt zu Grunde liegen, keiner Therapie. Sie sollten sich zurückbilden, sobald die ursächliche körperliche Einschränkung behoben ist.

Die Gangbilder von Patienten mit unterschiedlichsten Grunderkrankungen, z.B. Spastizität oder rein muskuloskeletalen Beeinträchtigungen, standen im Fokus dieser Arbeit. Das Ziel bestand darin, grundlegende Gesetzmäßigkeiten des krankhaften Gangs zu identifizieren. Diese Gesetzmäßigkeiten sollen in Zukunft die Unterscheidung zwischen primären und sekundären Gangbildabweichungen erleichtern. Mithilfe eines marker-basierten, dreidimensionalen Bewegungsanalyse-Systems (VICON) wurden Gangdaten der Patienten aufgezeichnet. Hiervon wurden hauptsächlich die kinematischen Gelenkwinkeldaten, elektromyographische Daten und Muskelkrafttests retrospektiv analysiert. Es wurde untersucht, ob und wie stark die Muskelkraft oder Orthesen Einfluss auf deren Gang haben, unabhängig von den Grunderkrankungen der Patienten.

Zunächst wurde eine Methode entwickelt und evaluiert, welche einen charakteristischen Gangzyklus für einen Patienten automatisch detektiert (SMaRT). Der Algorithmus basiert auf einer Hauptkomponenten-Analyse (PCA). Er ermittelt denjenigen Versuch, welcher über alle Gelenkwinkel-Kurven im Mittel am nächsten am Median aller Versuche liegt. SMaRT evaluiert die Daten objektiv und kann dabei eine uneingeschränkte Retest-Reliabilität aufweisen. Gleichzeitig ist die Methode mit 1,4 s/100 Daten-Sets um mehr als drei Größenordnungen schneller als die visuelle Identifizierung durch Experten. Besonders hervorzuheben ist die kleine Fehlerrate von 1,2% bei der Detektierung des representativen Trials. SMaRT kann sowohl durch anderen Gang-Parameter, z.B. Gelenk-Momente oder Gelenk-Leistung, erweitert werden als auch auf anderen Bewegungsanalyse-Kurven, wie z.B. in der sportartspezifischen Bewegungsanalyse, angewendet werden. Alle Studien in dieser Arbeit basieren auf dem repräsentativen Gangzyklus, welcher für jeden Patienten individuell durch SMaRT ermittelt wurde.

Für zwei weitere Studien wurden 716 Patienten anhand ihrer Erkrankungen in sieben Gruppen aufgeteilt: Orthopädische Patienten uni-/bilateral betroffen, neurologische Patienten uni-/bilateral betroffen mit hypotoner/spastischer Muskulatur mit/ohne Beeinträchtigung der Rumpfkontrolle. Ein negativer Effekt der Muskelkraft auf die Gelenkswinkel des Unterkörpers (Gait Profile Score, GPS) wurde mit der verallgemeinerten Methode der kleinsten Quadrate quantifiziert. Je schwächer die Patienten waren, umso stärker wich ihr GPS von der Norm nach oben ab. Die Stärke dieses Effekts unterschied sich in den sieben Patientengruppen nicht signifikant. Allerdings wurden zwischen den Gruppen bei einer normalen Muskelkraft signifikante GPS Unterschiede deutlich. Je höher der Schweregrad der Grunderkrankung, umso stärker war die Regressiongerade in Richtung eines höheren GPS parallel verschoben. Ortho-

pädische Patienten sowie Patienten mit zerebralparetischer Hemiplegie zeigten GPS-Werte, welche sich im Bereich derer von Gesunden bewegten (orthopädisch unilateral: 4.9°±0.7, orthopädisch bilateral: 5.0°±1.0, Hemiplegie: 5.4°±1.1). Dahingegen wichen Patienten mit Diplegie, Tetraplegie oder bilateral hypotoner Muskulatur signifikant von der Norm ab. Überraschender Weise wurde bei Patienten mit Diplegie und Patienten mit bilateral hypotoner Muskulatur eine gleich hohe GPS Abweichung von der Referenzgruppe beobachtet. Selbst der Vergleich der einzelnen Gelenks-Parameter zeigte nur geringfügige Unterschiede zwischen den beiden Gruppen.

Des Weiteren wurde eine Assoziation von Muskelkraft mit abnormer elektromyographischer Aktivität (EMG) gefunden, genauer gesagt mit verfrühter Plantarflexorenaktivität während der Gewichtsübernahme. Eine normale Muskelkraft reduzierte die verfrühte Gastrocnemiusaktivität um mehr als 10% über alle Patienten. Die Patientengruppe mit neurologischer Komponente und unilateral hypotoner Muskulatur stellte hier die einzige Ausnahme dar. Dies ist vermutlich auf die geringe Patientenzahl in dieser Gruppe zurückzuführen, welche eine Interpretation der Ergebnisse kaum zulässt. Auf Grund dessen, dass eine verfrühte Plantarflexorenaktivität in allen Patientengruppen auftrat, kann davon ausgegangen werden, dass diese Aktivität nicht nur durch die Grunderkrankung (z.B. Spastizität) hervorgerufen werden kann.

Abschließend wurde untersucht, ob eine Änderung in der Unterkörperkinematik eine Adaption im Oberkörper hervorruft. Hierzu wurden bei hemiplegischen Patienten die Oberkörperbewegungen beim Gehen auf Zehenspitzen (barfuß) und beim Gehen mit Fersen-Ballen Gang (mit Orthese) verglichen. Zwischen den beiden Konditionen wurden jedoch keine klinisch relevanten Unterschiede in der Rumpfbewegung gefunden. Allerdings verstärkte der gesunde Arm die Armschwungamplitude, um den reduzierten Armschwung der plegischen Seite zu kompensieren.

Schlussfolgernd kann festgehalten werden, dass die kinematische Änderung des sagittalen Sprunggelenkswinkels in den untersuchten Patienten nicht zu einer Normalisierung der Oberkörperbewegungen führten. Daher scheinen die verstärkten Oberkörperbewegungen nicht sekundäre Abweichungen zu sein, welche durch den Zehenspitzengang hervorgerufen werden. Anders verhalten sich hier die kinematischen Unterkörperabweichungen (Gait Profile Scores) und die verfrühte Plantarflexorenaktivität. Beide Abweichungen scheinen sekundär zu einer Muskelschwäche zu sein, was offenbar für alle Patientengruppen zutrifft. Während der Einfluss von Muskelkraft auf die untersuchten Gangparameter nicht unterschätzt werden darf, scheint die Spastizität das kinematische Gangbild geringfügiger zu beeinflussen. Bei der Interpretation von Ganganalysedaten sollte demnach die Muskelkraft immer beachtet werden, unabhängig davon welche Grunderkrankung der Patient aufweist.

SUMMARY

Walking for healthy humans is a basic everyday activity. In patients, especially those with neurologic disorders, the walking pattern can strongly deviate from those of healthy humans. For clinicians it is crucial to distinguish between primary and secondary gait deviations to plan the appropriate treatment. Primary gait deviations are causative and the target of therapy. Secondary gait deviations can be either active compensation or passive physical effects. Both do not require treatment as they are resolved when the primary impairment is corrected.

The focus of this thesis relied on the walking patterns of patients with different primary pathologies, e.g. patients with spasticity or orthopaedic patients. The aim was to identify principals of a pathological gait, with intention that it can further assist in differentiating between primary and secondary gait deviations. Three-dimensional gait analysis, driven by a marker-based movement analysis system (VICON) provided the data. All studies had a retrospective study design. Besides joint kinematics and surface electromyographic (EMG) data, muscle strength data were also analysed. It was investigated which parameters, such as muscle strength or orthotics, influences the gait of patients besides the primary pathology.

First, a method to detect a characteristic gait cycle for one subject was developed and evaluated. Based on Principal Component Analysis, the algorithm selects a trial that is closest to the median of all trials across different parameters, e.g. joint angle curves. The Selection Method for a Representative Trial (SMaRT) evaluates the data automatically, without subjective bias, and provides full repeatability. Furthermore, SMaRT required 1.4 s to analyse 100 datasets. Hereby, it was more than three orders of magnitude faster than the visual selection done by experts. Most importantly, the error rate of SMaRT with 1.2% was small; hence, the algorithm is relatively robust against a limited number of contaminated data. The algorithm can be individually adapted to any number and type of input parameters, e.g. joint moments, joint powers, etc. SMaRT is generally applicable to any type of curves derived by movement analysis, e.g. in the field of sports science. The representative trial for each subject was the base for further analysis.

To analyse the influence of muscle strength on gait deviations, patients were clustered into seven groups according to their pathology: orthopaedic patients uni-/bilateral, neurologic patients with uni-/bilateral flaccid/spastic muscles, with/ without thoracal control. The effect of muscle strength on the lower joint kinematics was calculated by generalised least squares. Muscle strength had a negative effect on gait kinematics, measured in the form of a Gait Profile Score (GPS). The weaker the patients were, the stronger the GPS differed from the norm. This effect was not significantly different across the different patient groups. Nevertheless, differences between the patients were found in the GPS offsets at normal muscle strength. The more severe the pathology of the patients, the higher the observed GPS offset was. Patients with orthopaedic diseases and patients with hemiplegic cerebral palsy were able to produce normal GPS values (orthopaedic unilateral: 4.9°±0.7, orthopaedic bilateral: 5.0°±1.0, hemiplegia: 5.4°±1.1). Patients with diplegia, tetraplegia, or flaccid muscles bilaterally differed significantly. Surprisingly, patients with diplegia and patients with flaccid muscles bilaterally showed the same GPS offsets to the reference group of 1.7° ±0.5. Even when comparing the particular joints, very few differences between these two groups were found.

Furthermore, muscle strength was observed to be associated with abnormal EMG timing, more exactly with premature plantarflexor activity during loading response of walking. Across all patient groups, a normal muscle strength reduced the number of patients with premature EMG by more than 10%. The only exception was the neurologic patient group with unilateral flaccid muscles. However the small amount of patients within this group might have biased the results. Abnormal EMG timing was prevalent across all pathology groups, indicating that it is not only caused by the primary pathology, e.g. spasticity.

Lastly, it was investigated whether a change in the gait kinematics of the lower body results in adaptations in the upper body. Hemiplegic cerebral palsy patients were compared when walking on their toes (barefoot condition) to a heel-toe gait condition evoked by wearing a hinged ankle-foot orthosis. No clinically relevant changes were observed in the trunk angle parameters when walking with orthoses compared to the barefoot condition. Nevertheless, the unaffected arm increased its swing amplitude and compensates for the reduced arm swing on the hemiplegic side.

In conclusion, kinematic changes of the sagittal ankle pattern in the specific hemiplegic patients do not result in a normalisation of the upper body kinematics. Consequently, none of the upper body abnormalities in this group seemed to be a secondary gait deviation resulting from toe walking. Contrarily, joint kinematics (GPS) of the lower body and activity timing of the plantarflexors can change secondary to the muscle weakness. Both effects seemed independent of the primary disease, at least to some extent. While the impact of muscle strength on the observed gait parameters cannot be neglected, spasticity seemed of minor importance. Therefore, muscle strength has to be taken into account when interpreting gait analysis data irrespective of the pathology.

CHAPTER 1

INTRODUCTION

HUMAN GAIT

Walking is one of the most basic human movements. According to Whittle [1] normal human gait is defined as "a method of locomotion involving the use of the two legs, alternately, to provide both support and propulsion" whereby "[…] at least one foot being in contact with the ground at all times". As bipedal walking might appeal natural and effortless to most healthy humans, some variability is present in normal gait. On the other hand, specific patterns of muscle activation as well as generated joint moments and powers can be identified in normal gait [1].

"Walking uses a repetitious sequence of limb motion to move the body forward while simultaneously maintaining stance stability" [2]. Normal gait is characterized by a stance phase, where the leg is loaded, supporting the body weight, and by a swing phase in which the leg is unloaded. These two gait sequences divide a gait cycle of 100% in a ratio of 60% to 40%. A gait cycle typically is defined as the period between two consecutive foot strikes of the same foot (0-100%) [2,3]. According to Perry [2], the stance phase can be subdivided into initial contact (IC) at 0%, loading response (LR) at 0-10%, mid stance (MSt) at 10-30%, terminal stance (TSt) at 30-50% and pre-swing (PSw) at 50-60% of the gait cycle. Likewise, the swing phase can be split into initial swing (ISw) at 60-73%, mid swing (MSw) at 73-87%, and terminal swing (TSw) at 87-100% of the gait cycle (Figure 1.1).

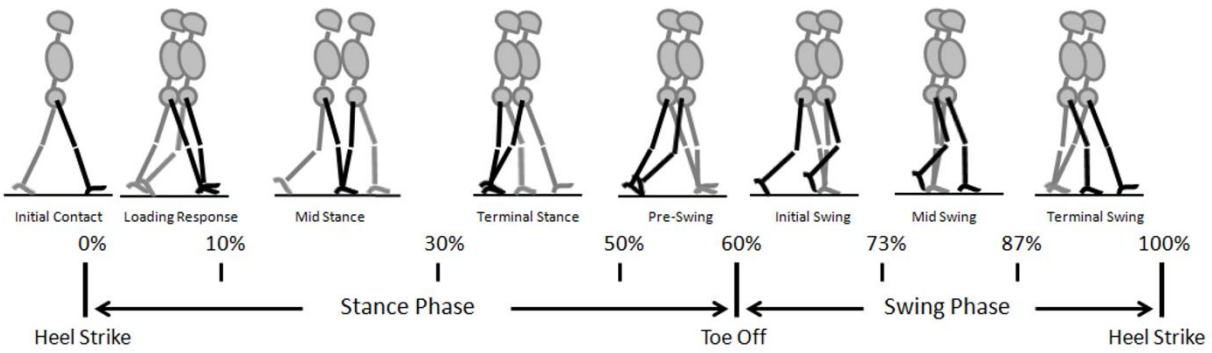

Figure 1.1: The gait cycle.
The gait phases of a gait cycle are illustrated according to Perry [2]. The stick figure shows the posture at the start and end of each gait phase, with the black leg as the reference leg.

Where healthy humans produce similar general walking patterns, patients with (neuro-)musculoskeletal impairment demonstrate a variety of gait deviations [2]. Their deformities and the dysfunction of the locomotor system require adaptations, which can be observed in their walking pattern. Pathological gait, can be more unstable, resulting in tripping, falling, or in a reduced gait velocity [4]. Further, it can be more energy consuming [5-7] and hence, limiting the walking performance of patients. Subsequently, the altered biomechanics during walking might lead to joint degeneration over the years [8,9]. For all of these reasons, the main focus of therapy in patients with locomotor dysfunction lies in the normalisation of

their gait pattern and/or preservation of their walking ability [10]. Here, one should not forget the psychological aspect of a normalised gait pattern. Some patients emphasise that they simply want to 'look more normal'. For the clinicians this means that the patient's gait deviations need to be identified in order to find suitable treatment options. Three-dimensional gait analysis provides valuable tools to assess human gait. "Clinical gait analysis allows the measurement and assessment of walking biomechanics, which facilitates the identification of abnormal characteristics and the recommendation of treatment alternatives" [11].

GAIT ASSESSMENT & GAIT ANALYSIS

Since the 80s "[…] gait analysis has been transformed from a purely academic discipline to a useful tool in the hands of physicians and therapists" [12]. The whole process of examining a patient's gait and making suggestions for treatment is termed 'Gait Assessment', while the term 'Gait Analysis' should be reserved to the technical side of the procedure [13]. Yet, it is a rather broad term, as it can include one or all of the following procedures: a detailed visual examination of the patient's gait, and/or quantitative measurements such as spatiotemporal parameters, joint angles, forces and electromyography (EMG) recordings [12,14,15]. While some gait abnormalities can be identified by eye, others can only be detected by using appropriate measurement systems [1]. In the following text passage an overview is given on the parameters assessed and analysed on behalf of this thesis. These parameters are: kinematics, kinetics, surface EMG, and clinical testing.

Kinematics

Marker-based systems are currently the state-of-the-art techniques in gait analysis [11]. They track the position of skin-mounted markers in a calibrated, three dimensional space. Typically three makers form a segment, which is simplified to a rigid body. On the basis of those data body segment movements in space, or in relation to each other, in terms of joint angles, can be calculated. Further the velocity and direction of the motion can be tracked [14]. These kinematic data are recorded and presented in three dimensions, namely in the sagittal, coronal, and transversal plane. One of those movement analysis systems (VICON, Oxford Metrics Limited, Oxford, UK) was used in our gait laboratory for data acquisition for this thesis. The Plug-in-Gait model [14], a conventional model in the field of clinical gait analysis [16-23], was applied for the kinematic calculations. The marker placement is defined in Table 1.1, and Figure 1.2 displays the markers and electrodes fixation on a patient. The joint angles of healthy subjects are presented in Figure 1.3.

Spatiotemporal parameters can easily be calculated from the marker trajectories. Walking velocity, step/stride time, step length/width, and the cadence grant a first impression about gait symmetry [2]. The cadence represents the number of steps per minute. According to Perry [2] healthy men have an average walking velocity of 1.43 m/s and women of 1.24 m/s. A normal stride length in adults is 1.4 m, a normal cadence is 120 steps per minute (2 steps/s) [1,24]. In children and adolescents these parameters vary along with age and height.

For scientific use, as in this thesis, the spatiotemporal parameters are commonly converted to non-dimensional values [25] to allow comparing subjects with different anthropometric appearance.

Table 1.1: Marker placement.
Listed are the Marker names and anatomical positions (placement) of the markers according to the Plug-in Gait model.

Marker name	Marker placement
LFHD / RFHD	Over the left/right temple
LBHD / RBHD	Back of the head left/right in a horizontal plane of the front head markers
C7	Cervical vertebra 7
T10	Thoracal vertebra 10
CLAV	Jugular notch where the clavicle meets the sternum
STRN	Xiphoid process of the sternum
LBAK	Left scapula
LSHO / RSHO	Left/right acromio-clavicular joint
LELB / RELB	Left/right lateral epicondyle
LWRA / RWRA	Left/right wrist bar thumb side
LWRB / RWRB	Left/right wrist bar pinkie side
LFIN / RFIN	Left/right below the head of the second metatarsal
SACR	Sacrum
LASI / RASI	Left/right anterior superior iliac spine
LTHI / RTHI	Left/right thigh in line with knee marker & trochanter major
LKNE / RKNE	Left/right lateral femoral epicondyles
LTIB / RTIB	Left/right shank in line with ankle & knee marker
LANK / RANK	Left/right lateral malleolus
LTOE / RTOE	Left/right second metatarsal head between fore-foot & mid-foot
LHEE / RHEE	Left/right calcaneaus same hight of toe markers

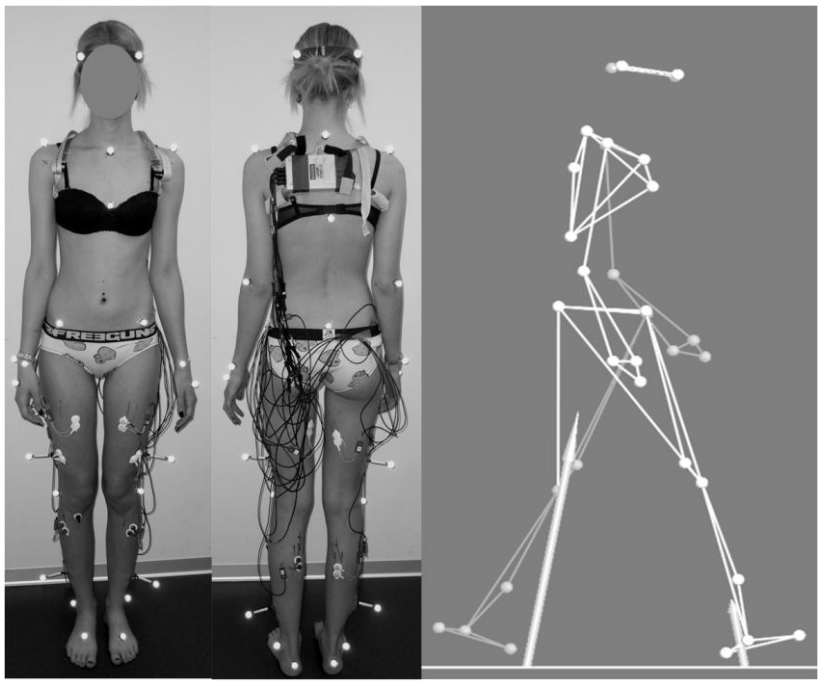

Figure 1.2: Marker and electrode fixation on a patient.
Figure A) shows the marker and electrode placement on a patient. The infra-red cameras capture the markers and joint them to body segments as illustrated in Figure B). The arrows depict the ground reaction forces measured by the force plates.

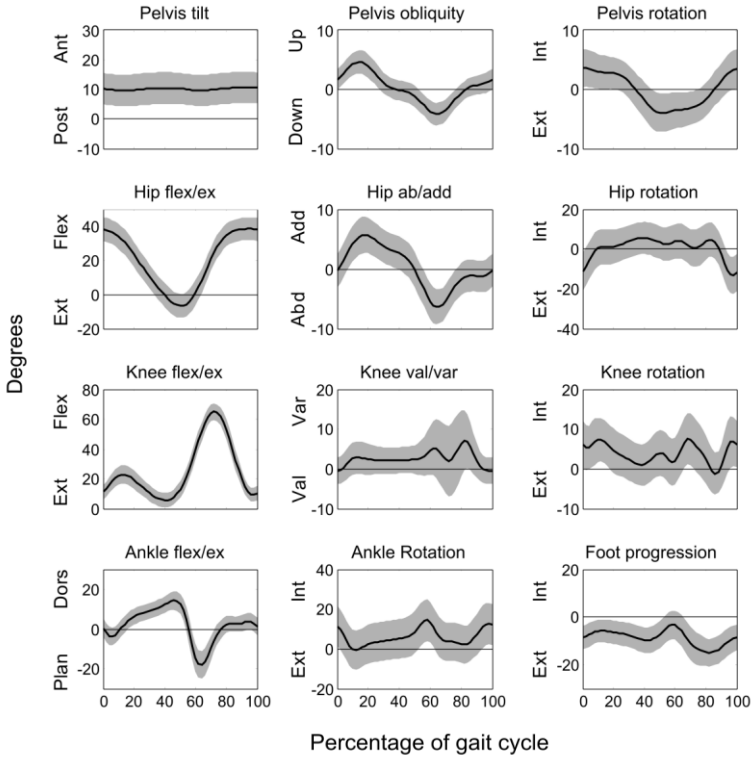

Figure 1.3: Joint angles of healthy subjects.
The mean and one standard deviation band of our healthy subjects are presented. The first column shows the angles in the sagittal plane, the second column presents the angles in the frontal plane, and the third in the transversal plane. The angles are time normalised to a gait cycle and are displayed in degrees. Post/ant = posterior/anterior, ext/int = external/internal, ext/flex = extension/flexion, add/abd = adduction/abduction, val/var = valgus/varus, plan/dors = plantar/dorsal.

Kinetics

The mechanical cause of movements are described through kinetics [15]. Force transducers measure kinetic or static dimensions of the movement. We have two force plates integrated into the floor of our gait laboratory which quantify the amplitude, the direction and the origin of ground reaction forces (GRF) while walking. The subjects have to encounter each force plate properly, with one foot only, to obtain feasable kinetic data. By means of 'inverse dynamics' one can calculate the joint moments and joint power (Figure 1.4), using the kinematic data together with the force plate output [12]. The segment masses, centres of gravity, and radii of gyration for each body segment are approximated according to anthropometric cadaver studies previously performed by Winter [26].

Joint moments are calculated as external moments created by the GRF. Internal moments are approximately equal but opposite to the external moments [3]. They result from muscle work and passive tissue resistance [27]. In gait analysis, the moment responsible for supporting the body against gravity typically is displayed as the positive moment, normalised to body mass (Nm/kg) [3]. Moments indicate which muscle group could be active, e.g. extensors or flexors. The joint power delivers the additional information whether this muscle group works eccentrically, absorbing energy, or generates power and therefore contracts concentrically [3].

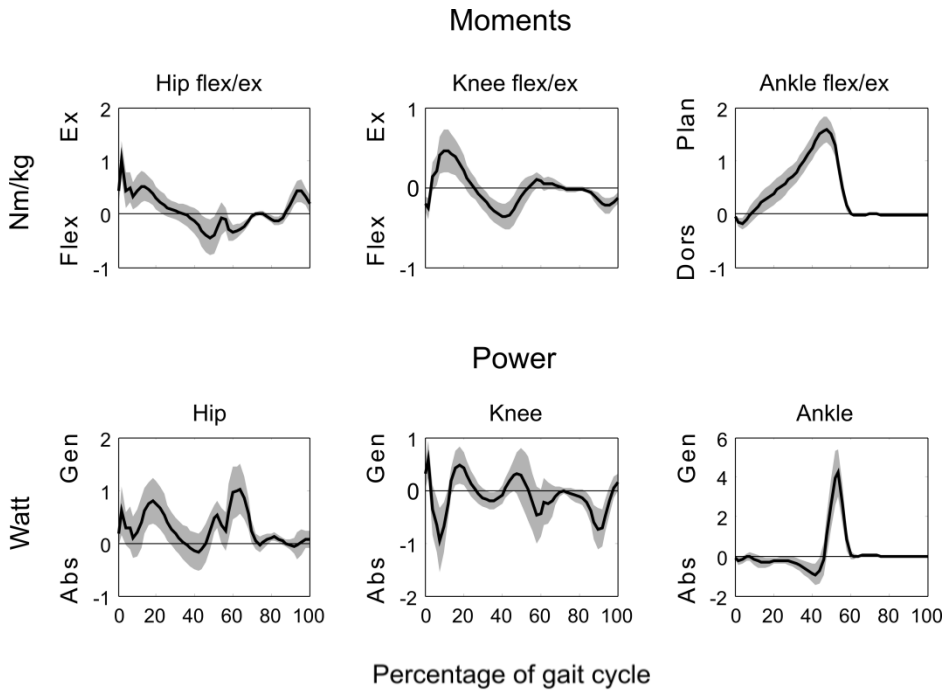

Figure 1.4: Kinetics of healthy subjects.
Presented are the mean and one standard deviation band of the kinetics in our normal subjects. The first row shows the joint moments in newton meter per kilogram body weight (Nm/kg). The second row shows the total power in watts. Both, moments and powers are time normalised to a gait cycle. Flex/ex = flexion/extension, dors/plan = dorsal/plantar, abs/gen = absorption/generation.

Electromyography (EMG)

Time-synchronised with the kinematic and kinetic data, surface EMG of the major muscles in the legs were recorded. EMG provides information on the neuromuscular activity. When a muscle changes its activation level, a temporal imbalance of ions around the muscle fibres originates [28]. This electrical potential difference is quantified by electrodes. In kinesiological studies mainly non-invasive surface electrodes are used. They are attached parallel to the fibre direction over the muscle belly [29,30]. In contrary to fine-wire EMG, surface EMG can assess muscles at the surface only.

EMG gives valuable information on the timing of muscle activity [31]. When interpreting secondary gait deviations, EMG assists in distinguishing between an active compensation and a passive effect. Interpretation of the EMG signals can either be based on the raw signal (Figure 1.5), as it is often the practice in a clinical environment, or on the processed signals. The on-off pattern of a muscle can be determined by a visual inspection of the raw signal [28,30,32]. A more quantitative analysis of the amplitude requires filtering [33], inverting and calculation of the mean trend, namely the envelop EMG [30]. By transforming the EMG signal to wavelets, information on timing and frequency of the signal can be interpreted [34].

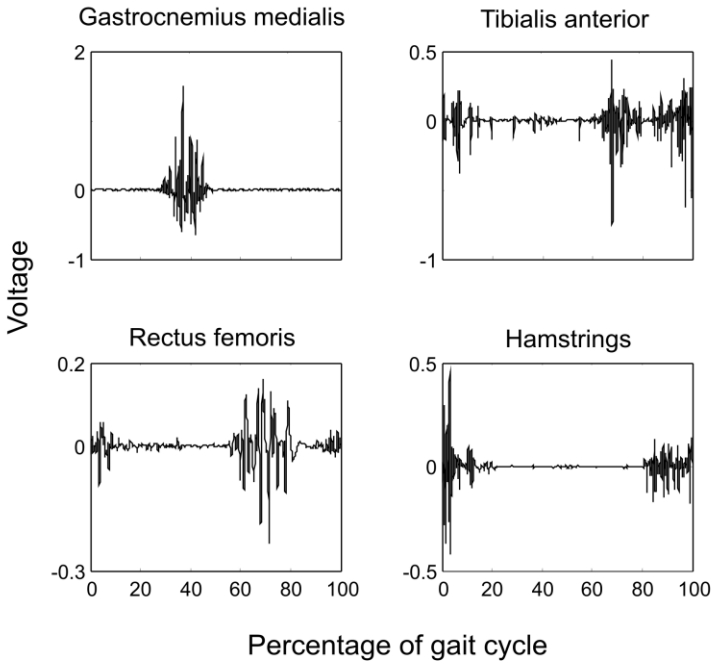

Figure 1.5: Electromyographic signal in healthy subjects.
The raw electromyographic signal (EMG) in one of our normal subjects is exemplified here. The four most important leg muscles for walking were selected. The signal is time normalised to a gait cycle and displayed in volts.

Clinical Testing

Together with the gait analysis, patients are commonly screened clinically. The physical examination can include a passive range of motion (RoM) evaluation of the joints, manual muscle strength testing [35], spasticity testing, and/or functional assessments [36].

The muscle strength was of interest for this thesis. It was manually determined for the hip flexors/extensors, abductors, internal/external rotators, the knee flexors/extensors, and the plantar flexors/dorsiflexors at the ankle. The scale ranges from 0 (muscle is paralysed) to 5, resembling maximum muscle strength [35]. As muscle strengths below 2 are not measurable without fine wire EMG, the physiotherapist determines the strength as follows:

<2 = no active movement possible
3 = active movement against gravity is possible,
4 = movement against a moderate restrain by the therapist is possible, and
5 = movement against hard restrain by the therapist is possible.

Often the clinical examination yields valuable information for the interpretation of the gait analysis data. For instance it is the case, if the knee flexion/extension angle derived from gait analysis shows that a patient is unable to extent his knee during walking. The RoM angle of the knee extension and the knee extensor strength will deliver the information as to whether the gait deviation is due to weakness or joint contracture.

PATIENTS

All studies included in this thesis were performed retrospectively. Gait data derived in daily clinical practice since 2001 were provided. The data were acquired in the Laboratory for Movement Analysis of the University Children's Hospital Basel (UKBB). Here, clinical gait analysis is conducted in patients with a wide variety of pathologies. In Table 1.2 the pathology distribution of the patients visiting in this specific laboratory is displayed. Merely patients who walked independently without walking aids were included. Similar to other clinical gait laboratories [11,37], the most prevalent group in our gait laboratory by far are children and adolescents with cerebral palsy (CP).

This thesis is not limited to a specific patient group; rather it includes patients suffering from various pathologies, stated in Table 1.2. Hence, only the main pathologies or disease groups can be briefly explained in the following. Furthermore, it is almost impossible to provide a complete overview on existing gait patterns in these patients. This is due to the following three reasons. Firstly, gait deviation can occur in various combinations for each individual patient, even given the fact that they suffer from the same disease. Secondly, gait patterns can change over time due to the development and growth of the children, especially in progressive pathologies. Lastly, as there exist simply too many gait deviations, describing them all is beyond the scope of this thesis. Therefore, only an excerpt of typical gait deviations in the following patient groups is described.

Table 1.2: Pathology distribution.
The pathology distribution in the Laboratory for Movement Analysis of the University Children's Hospital Basel (UKBB) between 1999 and 2012 is listed. Included in the statistics were freely, barefoot walking patients only (n=1131).

Type	Pathology	% of all patients	Absolute Nr.
Neuro	Hemiplegia (CP)	22.19%	251
Neuro	Diplegia (CP)	16.00%	181
Neuro	Tetraplegia (CP)	7.43%	84
Ortho	Torsional abnormality	5.48%	62
Ortho	Clubfoot	5.13%	58
Ortho	Leg length discrepancy	4.60%	52
Ortho	Patella dislocation	2.83%	32
Ortho	Other knee problems (e.g. fractures, total endoprothesis, tumors, pain, instability)	2.74%	31
Ortho	Talipes equinus (pes equinus)	2.74%	31
Neuro	Spina Bifida (Meningomyelocele)	2.56%	29
Neuro	Ataxia (CP)	2.21%	25
Neuro	Other neuromuscular diseases (e.g. Becker dystrophy, multiple sclerosis, myotonia, myopathy, HSMN, Polineuropathy, Myoclonic dystrophy (Curschmann-Steinert)	2.21%	25
Neuro	Paraplegia	2.21%	25
Ortho	Other ankle problems (e.g. fractures, total endoprothesis, tumors, pain, instability)	2.12%	24
Neuro	Developmental retardation / coordination disorder / unclear brain disorder	1.95%	22
Ortho	General disease with orthopead. problem (e.g. multiple osteochondromas, achondroplasia, pseudohypoparathyreoidism, dysmorphic syndrome,TAR-syndrome, Turner-syndrome)	1.41%	16
Neuro	Hemiplegia (not CP)	1.41%	16
Ortho	Knee ligament instability	1.33%	15
Ortho	Malalignement of knee axis	1.24%	14
Ortho	Planovalgus foot (pes planovalgus)	1.33%	15
Neuro	Poliomyelitis	1.24%	14
Ortho	Other hip problems (e.g. fractures, total endoprothesis, tumors, pain, instability)	1.06%	12
Ortho	Back pain	1.06%	12
Neuro	Nerve palsy (lower body)	1.06%	12
Ortho	Arthrogyposis multiplex congenita	0.88%	10

Ortho	Perthes disease	0.88%	10
Neuro	Duchenne's muscular dystrophy	0.88%	10
Ortho	Developmental dysplasia of hip (DDH)	0.71%	8
Ortho	Talipes calcaneus	0.53%	6
Ortho	Flatfoot (pes planus)	0.44%	5
Ortho	Scoliosis	0.44%	5
Ortho	Femoral deformity	0.27%	3
Ortho	Other spine deformity (not scoliosis)	0.27%	3
Ortho	Rectus fibrosis	0.27%	3
Neuro	Spastic hereditary paraparesis	0.27%	3
Neuro	Tetraplegia (not CP)	0.27%	3
Neuro	Down Syndrome	0.18%	2
Neuro	Spinal paralysis	0.18%	2

CP = cerebral palsy

Ortho = orthopaedic impairment

Neuro = neurological impairment

Cerebral Palsy (CP)

The term CP covers several symptoms caused by lesion of the central nervous system [38]. A uniform definition of CP does not exist. In their review, Kavčič et al. [39] list different definitions postulated throughout the years. There is a general consensus about the fact that CP is a non-progressive disease [38,40-45], but the movement characteristics and walking abilities of these patients can change throughout maturation [38,40]. CP is restricted to brain damage occurring prenatal or in early childhood [38,40,41,43-47]. In her surveillance of CP, Cans [38] states a prevalence of CP in Europe of 1.5-3 per 1000 live births.

The damage to the central control system causes some or all of the following abnormalities: "(1) loss of selective muscle control, (2) dependence on primitive reflex patterns for ambulation, (3) abnormal muscle tone, (4) relative imbalance between muscle agonists and antagonists across joints, and (5) deficient equilibrium reactions" [36]. Depending on their dysfunctions, patients can be either classified according to the topographical expression of the impairment, or according to the neurological implication. Topographically, the arm and leg of only one body side can be involved as in hemiplegic patients. The sensory and motor system of the contralateral side in these patients are usually relatively intact, and therefore walking is nearly always possible [36]. In diplegic patients, primarily both legs are affected. Most of those patients have the ability to walk, but they can show greater balance problems than hemiplegic patients and might need walking aids [36]. Tetraplegic patients have involvement of all four extremities [48]. Additionally, they suffer from a loss of trunk control. Some tetraplegic patients are still able to walk, but in many the balance and motor control is severely impaired.

Therefore, they are often unable to ambulate [36]. Alternatively, CP patients can be classified to three neurological implication groups: the group with spastic muscles, the group with ataxia, and the group with dyskinesia. Spastic CP is characterised by increased, not necessarily constant, muscle tone, and/or by pathologically increased reflexes. Movements of a patient with ataxia are performed with abnormal force, accuracy, and rhythm. Dyskinetic CP patients show involuntary, uncontrolled, recurring, and occasionally stereotypical movements. If the dyskinesia is dystonic, then the movements are stiff (hypokinesia) due to the increased muscle tone (hypertonia). Contrarily, choreo-athetotic dyskinetic CP is characterised by a reduced muscle tone (hypotonia) and stormy movements [38].

CP patients show a wide variety of gait deviations, depending on the age, severity, and anatomical location of their neurological impairment. Nonetheless, typical gait patterns are: equinus foot contact, in-toeing, and a stiff knee in swing. These gait deviations occurred in more than 50% of the hemiplegic, diplegic, and quadriplegic patients in the study of Wren et al. [49]. In the same study, more than half of the diplegic and quadriplegic patients presented increased hip flexion and crouch gait. Increased hip adduction was prevalent in more than 50% of the quadriplegic subjects [49].

Neurologic Patients with Flaccid Muscles

The problem of neurologic patients with flaccid muscles is a local or global muscle weakness. Typical pathologies that can result in local muscle weakness, where single muscles or parts of the body are flaccid, are Nerve Palsy or Poliomyelitis [50]. Contrarily, muscular dystrophies and Myelomeningocele globally weaken the lower/whole body, as can Poliomyelitis.

Myelomeningocele

Myelomeningocele (MMC) is the most severe form of Spina Bifida, where the neural tube is defective and spinal lesions occur [51]. In MMC patients, the vertebra was not fully closed in uterus, which can result in a protrusion of the spinal cord through the opening in the bones. This often causes muscle paresis and sensory defects proportional to the ascending vertebrae lesion level [21]. The most common gait deviations in MMC patients comprise of increased knee flexion due to plantarflexor weakness, excessive anterior pelvic tilt [52,53], and increased pelvic and trunk rotation and sway [53,54].

Muscular Dystrophy

Muscular dystrophy or myopathy are both umbrella terms for progressive muscle diseases, such as Duchenne's or Becker's muscular dystrophy. They all bring along structural and functional impairments of the skeletal muscles [55]. These impairments lead to muscle weaknesses, and consequently to constraints of the walking abilities [10,56,57]. Most myopathies implicate symmetrical extremity weakness, however, in some diseases the weakness can be asymmetrical [55]. With the progression of this disease, the weakness can cause the loss of ambulation [10].

Complementary to patients with neuromuscular pathologies, muscular dystrophic patients in general maintain preserved sensation [55].

The most common myopathy in early childhood is the Duchenne muscular dystrophy with 1 in 3500 live births worldwide. The disease is an X-linked recessive disorder. Children lose the ability to ambulate at a mean age of nine years [56]. A typical gait pattern observed in these patients is an excessively plantarflexed ankle during the swing phase and a compensatory hip flexion and abduction to maintain foot clearance [56].

Patients with Orthopaedic Diseases

Patients with an orthopaedic impairment typically show deformity, instability, or pain in their musculoskeletal system. A neurological cause for their impairment should be excluded. The impairment can involve only one joint on one leg, such as an instable knee due to a torn anterior cruciate ligament, or it can be symmetrical as it is commonly the case in habitual toe walkers. In our gait laboratory the majority of orthopaedic patients with isolated joint impairments have problems at the foot or knee. Hip impaired patients are relatively rare and thus not further described here.

Within the orthopaedic children/adolescents with foot problems seen in a gait laboratory, patients with clubfoot or habitual toe walkers are the most typical. This might be due to the fact that clubfoot occurs with 1 of 200 live births relative frequently. The foot of these patients is either plantarflexed or dorsiflexed at birth. The plantarflexed foot is inverted at the heel and forefoot, and adducted in the forefoot (talipes equinovarus). Contrarily, the talipes calcaneovalgus is dorsiflexed, abducted in the forefoot, and in eversion at the heel and forefoot. This deformity is in 50% of cases bilateral. Boys are twice as often affected as girls. Even when treated successfully, the foot can be smaller and less mobile than a healthy foot, which can result in functional problems during walking [58]. The foot is the only body segment that is in contact with the floor during the stance phase; hence, the foot acts as an effective lever arm to control the GRF. In patients with foot deformity where this lever arm is deformed (e.g. by clubfoot or toe walking) or instable (e.g. by mid-foot break), gait deviations can occur [59]. One study has revealed that a mid-foot break, together with an external rotation of the foot, leads to an internally rotated hip and pelvis [59]. In patients with clubfoot, the peak ankle plantarflexion moment was found to be reduced [60]. This indicates that the plantarflexion-knee extension couple is affected in these patients.

Patients with knee problems analysed in our gait laboratory mainly suffer from instable knees, such as torn knee ligaments or habitual patella dislocation. The knee is the most critical joint of the lower body to control due to its anatomical structure. Without a bony enclosure, this joint is primarily stabilised by muscles, ligaments and surrounding tissues [61]. When those structures are injured or lax, a patient can show a reduction in the magnitude of the flexion moment about the knee to avoid or reduce the contraction of the quadriceps [62].

GAIT DEVIATIONS

While healthy people can have walking patterns with similar kinematic, kinetic, and EMG parameters [63], these parameters can deviate strongly in patients with (neuro-)musculoskeletal diseases [2]. Gait deviations can be either primary or secondary. *Primary gait deviations* are defined as a direct result of the pathology [17]. For instance, patients with weak hip abductors can show a pelvic drop of the contralateral side in single stance, namely the Trendelenburg sign [24]. In these patients, the ipsilateral hip abductors are too weak to hold the pelvic up against the weight of the upper body when the contralateral leg is in the swing phase. Patients with spastic contractures of the hip adductors can also show signs of Trendelenburg. In these patients, the ipsilateral contracted hip adductors pull the pelvis down on the contralateral side [24].

Secondary gait deviations have to be divided into passive physical effects and active compensations. Passive effects follow as a physical consequence of the primary deviations [64]. When the biomechanics of one joint is altered as a result of the pathology, then anatomical coupling of the body parts and gravity acting on them inevitably result in deviations of the other joints. Forward simulation has revealed that when the leg is loaded during the contracture of the triceps surae muscle, hip flexion, internal rotation, and adduction together with external pelvic rotation [65] is produced. This computer model has no ability for active interaction; hence, the physical effects are of purely passive nature. Contrarily, active secondary deviations, or compensations, work in order to actively offset the primary deviations and secondary physical effects [17,64,66-68]. These compensations are needed to maintain adequate functionality. For example, in a patient with a strong sign of Trendelenburg, the pelvis was positioned too low, giving the swinging leg not enough space to swing through. In order to gain foot clearance the thorax can be actively shifted laterally over the stance limb (Duchenne gait). As the pelvis is connected to the upper body, this will pull the contralateral pelvis up, resulting in an enhanced foot clearance [24].

Both active and passive secondary deviations are the main topic of this thesis. Therefore, an overview of active compensations and passive physical effects described in the literature is provided in the following.

Secondary Passive Physical Effects

As described previously, excessive plantarflexor work that primarily leads to an equinus ankle position provokes secondary hip flexion, internal rotation, and adduction together with external pelvic rotation [65]. In the past, these passive effects had been incorrectly referred to as compensations [69,70]. Excessive foot rotation during stance, resulting in increased hip rotation, is another passive effect. When an abducted (externally rotated), plano-valgus foot is dorsiflexed under loading conditions, the tibia is automatically pushed into an internally rotated position. This distant effect of the foot rotates the entire leg internally [59]. Therefore, the rotation of the leg is seen as an internal rotation at the hip and pelvis. Likewise, an internally rotated foot (in-toeing gait) produces an external rotation at the hip [16,71,72]. In Table 1.3, secon-

dary physical effects are listed of patients which were observed in laboratory settings, together with the computer models that were used in studies.

Table 1.3: Physical effects.
The table summarises the passive physical effects identified in the literature. This table is a fusion and amendment of tables 3 & 5 in Schmid et al. [64].

Biomechanical constraints due to primary pathology	Physical effect	Patients showing this effect
Equinus position of ankle (toe walking)	Anterior pelvic tilt (hip flexion), hip internal rotation and external pelvic rotation (pelvic retraction)	Spastic hemiplegic CP [17,69,70]; Spastic diplegic CP [67,70]; 3D full body forward simulation [a] [65]
Internal rotation deformity of foot (in-toeing gait)	Hip external rotation *	Idiopathic clubfoot [16,71]; Charcot-Marie-Tooth disease, types I and II [72]
External foot rotation due to 'mid-foot break'	Hip/pelvic internal rotation	Diplegic cerebral palsy [59]

* indicates effects that appear to be independent from the underlying pathology.

[a] computer simulation studies

CP = cerebral palsy

Secondary Active Compensations

The walking ability of patients is mainly restricted by a reduced RoM in selective joints or by a weakness of muscles [15].

Compensations for Muscle Weakness

Patients generally have three main principles to compensate for muscle weakness. First, they can use synergistic muscles to replace or support the weak muscles. Second, they can displace the centre of mass (CoM) to reduce the GRF arm at a joint. Third, they can 'restore' the lever arm of a specific muscle group.

Muscular weakness in one muscle or muscle group can be counter-balanced by synergistic muscles. When plantarflexors are weak, then different compensations can be applied. The forward propulsion can be sustained by higher hip and knee extension moments in stance phase [6,60,73]. Prolonged EMG activity in the vastus medialis and rectus femoris muscle was found to support these mechanisms in patients [6]. In a modelling study, a prolonged hamstring contribution to the support moment was needed when the soleus contribution was diminished [74]. Furthermore, patients with chronic stroke can have weak plantarflexors, which

limits push-off in these patients. Hip extensor strength was reported to be positively associated with walking speed within stroke patients [75]. The hip extensor torque in late stance was supposed to 'load' the passive hip flexor structures that can 'pull' the leg up during toe-off [75]. The hip flexors were also described to actively pull the leg up to achieve foot clearance in patients [6,76] and in a musculoskeletal modelling study [73]. Riad et al. [77] observed a larger power generation at the hip to balance a decreased ankle power generation in hemiplegic CP patients. Further, co-contraction of the hamstring and quadriceps muscles can be used to reduce the net quadriceps moment at the knee [62,78-80]. In patients with an instable or painful knee quadriceps femoris muscle activity can cause an increased tibia forward translation. The hamstrings can hold the tibia back. The co-contraction around the knee was found to stabilise this joint by shifting the hamstrings activity to extend the hip instead of flexing the knee [32]. This mechanism is supported by a single case modelling study where one patient substituted the knee extensor moment by an increased hip extensor moment [81].

A global leg weakness can be compensated by hyperactivity of the ankle plantar flexors around foot strike. This activity controlled the leg of weak orthopaedic patients [32] by the plantarflexion-knee extension couple [36]. The authors of the same study proposed that the co-contraction of the knee extensors and hamstrings produces a shift of the hamstrings activity from a potential knee flexion to a hip extension [32]. A prolonged activity of the contralateral hip abductors was found to decelerate the weight acceptance on the ipsilateral limb [82]. This mechanism reduces the angular velocities, and thereby, the flexion moments one has to counteract during loading response. Van der Krogt et al. [83] simulated muscle weakness in a forward modelling study. They systematically reduced the force applied by the muscles of the legs in their model. Then, they analysed which synergistic muscles increased their activation to compensate for the weakened muscle. Further, it was evaluated whether the activation in the weakened muscle increased, and which of the antagonistic muscles decreased their activation in response to the weakness. Table 1.4 lists the main results. Their results were supported by Knarr et al. [84], who described the plantarflexors and hamstrings to compensate for each other. Additionally, Jonkers et al. [85] confirmed the hamstrings to contribute to stance hip extension when the gluteus maximus muscle is weakened. The same authors also reported the gastrocnemius and soleus muscle to compensate for each other in their muscle model.

External flexion/extension moments are created by the GRF arm. For instance, the GRF tends to dorsiflex the ankle in a standing position, as the GRF is anterior to the ankle. These external moments must be counterbalanced by an internal moment created by muscles and passive structures on the opposite side of the joint [36]. In the example this would be the Achilles tendon and the triceps surae. By translating the CoM so that the GRF vector moves closer to the joint centre or even on the opposite side of the joint, one can reduce the muscle work needed to stabilise a joint. Therefore, patients with weak hip extensors were found to extend the trunk posterior to shift the CoM behind the hip in order to produce an external hip extension moment [53,86]. Hip abductor weakness was found to be handled by trunk lean over the affected leg in single leg stance, namely in patients with Duchenne limp [21,53,87-89]. Patients with knee extensor weakness translated the CoM anterior by forward trunk lean. This was achieved either by flexion of the hip [90-93] or anterior pelvic tilt [94]. As a result the external knee flexion

moment is changed into an external extension moments and the use of the quadriceps can be avoided. The activation of the plantarflexion-knee extension couple has a similar effect [32,73,81,82,95,96]. The plantarflexor activity at the ankle during loading response and mid stance displaces the centre of pressure forward along the foot. The GRF that is posterior to the knee in these gait phases is, thereby, transferred closer to the knee joint centre [82].

Table 1.4: Compensations to simulated muscle weakness.
The table shows the compensations by the same muscle, by synergistic muscles, and by antagonistic muscles as a reaction to a specific, weakened muscle. The table is a modified version of Table 1 in Van der Krogt et al. [83]. Abbreviations: GMAX: gluteus maximus, GMED: gluteus medius, ILPS: iliopsoas, HAM: hamstrings (semitendinosus, semimembranosus and biceps femoris long head), RF: rectus femoris, VAS: vasti (vastus medialis, lateralis, and intermedius), TA: tibialis anterior, GAS: gastrocnemius (medialis and lateralis combined), SO: soleus, GMIN: gluteus minimus, QF: quadratus femoris, PIRI: piriformis, SMM: semimembranosus, TFL: tensor fascia lata, BFS: biceps femoris short head, SAR: sartorius, GRA: gracilis, ADD: adductors, PERT: peroneus tertius, EXTD: extensor digitorum longus, TIBP: tibialis posterior, FLD: flexor digitorum longus, FLH: flexor hallucis longus, PERB: peroneus brevis, PERL: peroneus longus, SMT: semitendinosus, PSO: psoas, (sw): only in swing. Muscles in brackets only have minor contributions. More ventral parts are indicated by lower numbers (GMAX1, GMED1, etc.), dorsal parts of the muscle by higher numbers.

Weakened muscle	Compensations		
	Increased activation in this muscle	Synergistic muscles that increase their activation	Reduced activation in antagonistic muscles
GMAX	-	GMED3 (GMED2 GMIN3 HAM ADD VAS QF PIRI)	-
GMED	✓	GMIN SMM TFL BFS SAR GMAX1 GAS RF (VAS)	PSO GMAX2,3 SO ILPS Up
HAM	✓	SAR GRA ADD GMAX GAS TA PERT EXTD	GMED2,3 SO
RF	✓ (sw)	ILPS VAS SO TFL (GMED2,3)	SMT BFS GAS GRAC TA
VAS	✓	ADD GMAX GMIN1	HAM (sw) ILPS (sw)
TA	-	EXTD PERT	-
GAS	-	SO BFS SMT SMM ILPS (GMED GMIN SAR)	TA
SO	-	GAS TIBP FLD FLH PERB PERL VAS RF	TA EXTD BFS ILPS SAR (GMIN)

✓ marks increased activity

- indicates no increased activity

Lastly, the muscle's force generating capacity can be diminished by reduced force arms due to deformity. In patients with increased femoral anteversion the force arm of the gluteus medius muscle was found to be impaired. For this reason, these patients suffer from hip abductor weakness. In order to restore the moment arm of the gluteus medius they produce an internal rotation at the hip [97]. Contrarily, patients with an increased Q-angle showed a reduced internal rotation of the hip [98]. The Q-angle is the angle between the elongated tibia and the line from the mid-point patella to the anterior superior illiac spine. Reducing the internal rotation of the femur could ease the lateral force vector on the patella, and restore the force arm of the quadriceps muscle [98]. Aside from that, the outward spreading of the arms can be a compensation to keep balance during walking in patients with diplegic CP [99,100].

Compensations for Restricted Range of Motion

Restricted range of motion in a joint can be compensated by increased motion in adherent joints. A limited hip extension that would lead to a trunk forward lean, was adjusted by an increase lumbar lordosis [87] or knee flexion [101] to keep the trunk near the vertical. Similarly, a loss of lumbar lordosis would move the centre of mass anterior. This was prevented by hyper-extending the hips [102] or flexing the hip and knee and dorsiflexing the ankle (crouch gait) [102,103]. The same three joints were flexed to functionally shorten the longer leg in patients with leg length discrepancy [104]. Patients with a reduced foot clearance during swing phase of walking were found to have five different compensatory patterns: three on the sound side and two on the affected side. The sound side was functionally elongated by lifting the heel from the floor (vaulting) while the contralateral leg was in swing [17,77,92,105,106]. Contraction of the hip abductors on the unaffected side leveraged the pelvic up on the problematic side (pelvic hike) to gain foot clearance [6,53,75,92,107]. Excessive compensatory posterior pelvic tilt while standing on the sound leg was reported to accelerate the affected foot into swing [2,76]. The affected leg was also seen to be swung in a half-cycle around the stance leg (circumduction) typically together with excessive hip abduction and external rotation [6,56,72,91,92]. Increased hip and/or knee flexion in form of a steppage gait was found to lift the affected leg of the floor. All five compensatory patterns can be combined with each other. Table 1.5 provides a comprehensive overview on described gait compensations in the literature. Further, it informs about in which pathologies they were observed or the computer models that were used in the studies.

Table 1.5: Compensatory mechanisms.
The compensatory mechanisms known from the literature are summarised in the second column. The patients or conditions under which they have been observed (e.g. single case study, simulation study) are listed in the third column. This table is a fusion and amendment of tables 3 & 4 in Schmid et al. [64].

Constraints due to pathology	Compensatory mechanisms	Patients showing this compensation
Hip extensor weakness	Posterior trunk extension	Myelomeningocele [53]; facioscapulohumeral muscular dystrophy [c] [86]
Hip abductor weakness	Duchenne limp *	Myelomeningocele [21,53]; Unilateral osteoarthritis hip [87]; Legg Calvé Perthes disease [88]; Hemi- & diplegic CP [89]; three-dimensional model (3D) [a] [93]
	Restoring moment arm by internal rotation	Patients with excessive femoral anteversion [a] [97]
Knee extensor weakness / Quadriceps avoidance	Hip extensors (hamstrings) for knee extensors *	Unilateral anterior cruciate ligament deficiency [62,78,79]; Several orthopedic conditions [32]; Chronic patellofemoral pain syndrome [108]; Two dimensional (2D) model of the knee [a] [109,110]; Full body 2D musculoskeletal model [a] [73]; 3 patients with knee extensor weakness due to sarcoma or amyotrophic lateral sclerosis [c] [81]
	Centre of mass anterior to the knee joint by:	Several orthopaedic conditions [32]; Hereditary spastic paraplegia & mild spastic diplegia CP [95]; Spinal muscular atrophy, type III [82]; Full body 2D musculoskeletal model [73,96] [a]; 3 patients with knee extensor weakness due to sarcoma or amyotrophic lateral sclerosis [c] [81]
	Increased activity of plantarflexion-knee extension couple *	
	Hip flexion *	Juvenile chronic arthritis [90]; Hemiparesis after stroke [91,92]; 3D model [a] [93]
	Anterior pelvic tilt	Bilateral, medial osteoarthritis of the knee [94]
	Kneehyperextension (e.g. by prolonged contralateral plantarflexor activity)	Duchenne muscular dystrophy [60]; 3 patients with knee extensor weakness due to sarcoma or amyotrophic lateral sclerosis [c] [81]
Ankle plantarflexor weakness	Eccentric work of hip flexors for progression in stance	Unilateral congenital clubfoot [60]
	Hip and knee extensors in stance *	Unilateral congenital clubfoot [60]; Charcot-Marie-Tooth disease [6]; Full body 2D musculoskeletal model [a] [73]; 3D full body model [a] [111]
	Hip flexors (pulling) in preswing *	Charcot-Marie-Tooth disease [6]; Hemiplegia after stroke [76]; Full body 2D musculoskeletal model [a] [73]
	Hip extensor torque strategy in late stance (loading flexor tissue)	Chronic stroke [75]; 2D full body model [a] [74]

	Internal rotation of trunk and pelvis on contralateral side	Myelomeningocele [53]
	Larger symmetrical hip power generation	Spastic hemiplegic CP [77]
Global leg weakness	Hyperactivity ankle plantarflexors *	Several orthopaedic conditions [32]; Spinal muscular atrophy, type III [82]; Full body 2D musculoskeletal model [73] a
	Co-contraction around knee	Several orthopaedic conditions [32]
	Prolonged activity of contralateral hip abductors (weight acceptance)	Spinal muscular atrophy, type III [82]
Gait instabiliy	'Guard position' of the arms (increased abduction in shoulder & elbow flexion)	Spastic diplegic CP [99,100]
Limited hip extension	Lumbar lordosis	Unilateral hip osteoarthritis [87]
	Knee flexion to allow the pelvis to progress forward	Unilateral congenital dysplasia of the hip [101]
Loss of lumbar lordosis (Center of mass anterior)	Hip hyperextension	Postoperative flatback [102]
	Crouch gait	Postoperative flatback [102]; healthy subjects asked to walk with flexed hip b [103]
Rotational knee instability / increased medial knee load	Lateral shift of center of mass (e.g. pelvic hike) *	Anterior cruciate ligament deficiency [78]; Bilateral medial knee osteoarthritis [8,94]; Healthy subjects walking with increased medio-lateral trunk sway b [112]
Patella 'out of line' (Q-angle increased)	Reduced hip internal rotation	Patellofemoral pain syndrome [98]
Knee pain	Co-contraction of quadriceps and hamstrings	Patellofemoral pain, patient driven model a [113]
Leg length discrepancy	Hip, knee flexion and ankle dorsiflexion on unaffected (longer) side	Spastic hemiplegic CP with leg-length discrepancy [104]; artificial long leg by raising the sole of one foot using pelite b [114]
Reduced foot clearance	Pelvic up tilt (posterior tilt) on unaffected side	Hemiplegia after stroke [2,76]
	Pelvic hike *	Chronic stroke [75]; Charcot-Marie-Tooth disease [6]; Myelomeningocele [53]; Hemiplegia after stroke [92,107]; Artificial unilateral knee immobilization b [115,116]
	Circumduction, hip abduction, hip external rotation *	Duchenne muscular dystrophy [56]; Charcot-Marie-Tooth disease [6,72]; Hemiparesis after stroke [91]; Hemiplegia after stroke [92]
	Hip flexion and/or knee flexion (steppage gait) *	Duchenne muscular dystrophy [56]; Charcot-Marie-Tooth disease type 1A [105]; Unilateral artificial reduced knee flexion in healthy subjects b [115]

	Increased plantarflexion on unaffected side (vaulting) *	Charcot-Marie-Tooth disease type 1A [105]; Spastic hemiplegic CP [17,77]; Hemiplegia after stroke [92]; Artificial unilateral knee immobilization [b] [115,116]; Traumatic brain injury [c] [106]
Initial toe contact	Early onset of plantarflexors, reduces dorsiflexor activity	Hemiplegic CP [117]

* indicates compensations that appear to be independent from the underlying pathology.

[a] computer simulation studies

[b] in-vivo simulation studies / mimicking studies

[c] single-case studies

CP = cerebral palsy

RELEVANCE & AIMS

Clinical gait analysis is primarily used for treatment planning in patients. For the clinician it is hereby essential to identify the gait deviations that are primary and those which are secondary [1]. This differentiation is crucial, as only primary gait deviations should be targeted by medical treatment. As soon as the primary source of the problem is corrected, the secondary gait deviations are meant to resolve spontaneously [17,68]. When secondary deviations are mistaken as cause of the gait abnormality and treated accordingly, this therapy can either be inefficient or even deteriorate the walking performance of a patient [17,66-68]. "Regrettably, errors of this type are all too common, particularly when treatment is prescribed without the benefit of gait assessment" [1]. Unfortunately, the distinction between primary and secondary gait abnormalities is not always obvious. Especially an overuse of the term 'compensation' is present in literature [64].

Comparing a pathological gait pattern to a healthy one, as it is common practice, does not allow a clear differentiation between primary and secondary gait deviations [64]. A more suitable method is to investigate patients under two different conditions, e.g. pre and post surgery [17], or with and without orthotics. Those gait parameters on the other joints that become normal after surgery, or with orthotics, are most likely secondary gait deviations. Another possible method is to have healthy controls mimicking the gait pattern of a specific patient group, such as toe walking [117]. Similarly, in vivo simulation studies, where a primary abnormality is induced can help to reveal secondary abnormalities [64]. Walsh et al. [114] artificially elongated one leg of healthy subjects to simulate leg length discrepancy [114]. Short hamstrings were imitated by a knee brace limiting knee extension by Whitehead et al. [118]. Computer simulation [73,74,80], and especially forward modelling [65], are useful tools to distinguish between physical effects and compensations. In forward modelling it is possible to change one parameter, e.g. excessive plantarflexion, and evaluate if this alteration has a physical effect on adherent joints [65]. Otherwise, one can search for similar gait patterns in patients with various pathologies. If patients with different primary diseases show similar gait deviations, these alterations are most likely secondary. Literature on such studies is scarce. To

the authors knowledge only one study has assessed abnormal muscle activity in patients with different orthopaedic impairments [32]. However, Table 1.5 implies that there are compensations that are independent of the primary pathology.

For all the reasons stated above, the objective of this thesis was to identify principles of pathological gait that are independent of the primary pathology. For instance, some gait deviation could be a result of the (neuro-)musculoskeletal alteration in the first place, e.g. muscle weakness or joint stiffness, rather than spasticity. Therefore, it was investigated how muscles strength and changes of the gait pattern, e.g. when wearing orthoses, do influence the gait deviations of patients. The research question was whether there are similarities in the gait patterns of patients with various primary diseases. It was aimed to provide an overview on the association between the muscle strength in relation to gait kinematics and abnormal EMG patterns during walking in patients with different (neuro-)musculoskeletal diseases. The hypotheses were: (i) A negative correlation between the amount of gait deviation and the mean manual muscle strength of the leg muscles exists; (ii) This correlation is similar across different patient groups; (iii) The severity of the pathology is reflected in a higher gait deviation in patients with normal muscle strength; (iv) Abnormal EMG activity is present in all patient groups; (v) Muscle weakness and equinus gait are aetiological factors for EMG activity independent of the pathology. Furthermore, the effect of a modified walking pattern of the lower limbs on the gait deviations of the upper body was evaluated. This was achieved by comparing hemiplegic CP patients when walking on their toes (barefoot) and when an orthosis corrects their ankle pattern to a heel-toe gait.

ANALYSIS METHODS

Principal Component Analysis (PCA)

The Principal Component Analysis (PCA) is a common method for dimensionality reduction of high dimensional data. Mathematically, the PCA converts the i inter-related variables $X = x_1, x_2, \ldots, x_i$ with an orthogonal transformation into a mutually uncorrelated space. The principal component vectors (PC-vectors) are the eigenvectors $E = e_1, e_2, \ldots, e_i$ of the covariance matrix of X, and they are often arranged in decreasing order of their sample variances. Hence, the first eigenvector is where the highest variance of the data is found and so on. $Z = z_1, z_2, \ldots, z_i$ is the principal component score (PC-score) vector derived from the product of the eigenvectors and the data, hence $Z = E^T X$, with the variables z_i referred to as PC-scores. They contain information about the contribution of the PC-vector to the individual waveform [119].

In the field of human movement analysis PCA can be successfully applied as feature extractor or as a data-driven filter [120]. Due to the sensitivity of a PCA to the waveform [121], and as a data reduction technique [119,122], PCA became a valuable tool used on time series data such as joint angles in biomechanics [123-127]. In this thesis PCA was applied to kinematic gait data to select a representative trial (see *Chapter 3*).

OUTLINE

This cumulative thesis includes four publications addressing the two general research questions: Which gait deviations are primary and which are secondary, and can similar gait deviations be observed in patients with different pathologies? The manuscript in *Chapter 2* provides a method to analyse gait data. *Chapter 3* and *4* consist of two cross-sectional studies comparing patients with different pathologies regarding their similarities and dissimilarities in gait. In *Chapter 5* the effects of hemiplegic toe walking on the upper body were investigated.

Chapter 2 describes a method based on PCA [119] to find a representative trial among numerous measurements of a patient. Each patient, undergoing a gait analysis in the laboratory, has to walk several times under the same condition. Thereby, it is assured that one obtains a few characteristic trials or gait cycles for each patient. In order to be able to compare gait between different patients or between normal subjects and patients, one trial per person had to be selected. The aim of this study, therefore, was to develop an algorithm for an automatic detection of a representative trial. The developed algorithm is described in detail and the results of its evaluation are presented and discussed.

Chapter 3 evaluates how the muscle strength influences the gait kinematics in patients with different pathologies. The gait analysis data of 716 patients were retrospectively assessed. All patients were clustered into seven patient groups. The groups were formed according to the source of the problem: orthopaedic, neurologic with flaccid or spastic muscles, with or without trunk control, and uni- or bilaterally involved. The Gait Profile Score [128] was calculated from the joint angles of these patients in comparison to healthy controls, as a global measure on the quantity of gait deviation. Manual muscle strength testing [35] delivered the mean muscle strength of each patient. By means of the generalised least square models, the correlation between muscle strength and Gait Profile Score was calculated. Additionally, it was tested whether the influence of muscle strength on the gait pattern defers between the different patient groups.

Chapter 4 addresses premature plantarflexor activity in the loading response during walking. Throughout this gait phase the calf muscle is typically quiet within healthy subjects. Two hypotheses where explored retrospectively: (i) premature plantarflexor activity correlates with equinus foot contact, and (ii) weak patients show premature gastrocnemius muscle activity more often than patients with normal muscle strength. This study provides also an overview on the prevalence of premature gastrocnemius activity in the same seven patient groups as in *Chapter 2*. To avoid a possible bias by the patient group clustering according to their diagnosis, all patients were also clustered according to their impaired joints. The hypothesis (ii) was tested again on this second patient clustering.

Chapter 5 discusses the effects of toe walking and hinged ankle-foot orthoses (hAFO) on the upper body kinematics of hemiplegic CP patients. These patients typically walk on their toes on the hemiplegic side. Therefore, it is possible that some of the upper body deviations that are clinically observed are rather secondary to their instable foot position than primary due to spasticity. In a first step, it was detected which upper body joint angles deviated from the

norm when the patients walked on their toes. In a second step, the same patients were analysed while walking with an hAFO, which corrected their foot position to a heel initial contact. The main objective was to investigate if any of the abnormal upper body parameters while toe walking are corrected by wearing an hAFO.

The thesis is completed by a conclusion and outlook on future research in *Chapter 6*.

CONTRIBUTORS

In order to secure the best possible outcome, the contents of the following chapters were developed within an interdisciplinary team. Although the main work was done by the author of this thesis (KS), five experienced scientists have supplied valuable contributions, namely, Prof. Dr. med. Reinald Brunner (RB), Dr. Jacqueline Romkes (JR), Prof. Dr. Bert Müller (BM), Prof. Dr. Philippe Cattin (PCC), Dr. Cora Huber (CH), and Dr. Michael Coslovsky (MC). In the following the contributions of the authors are listed. The authors' order is the same as in the published articles.

Chapter 2: A Selection Method for a Representative Trial

PCC: Assistance with development of the algorithm, and critical reviewing of the manuscript for important intellectual content.

RB: Assistance with development of the algorithm, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

BM: Suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

CH: Assistance with programming the algorithm, evaluation of the algorithm, and critical reviewing of the manuscript for important intellectual content.

JR: Assistance with development of the algorithm, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

Chapter 3: The Influence of Muscle Strength on Gait Kinematics

JR: Design of the study, suggestions for data interpretation, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

MC: Statistical calculations, and assistance with writing of the results and discussion section.

RB: Design of the study, suggestions for data interpretation, suggestions for graphical representation of figures, drafting of the introduction and discussion section of the manuscript, and critical reviewing of the methods and results section for important intellectual content.

Chapter 4: The Influence of Muscle Strength and Equinus Gait on EMG

JR: Design of the study, suggestions for data interpretation, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

RB: Design of the study, suggestions for data interpretation, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

Chapter 5: The Effect of Toe Walking on the Upper Body

RB: Design of the study, suggestions for data interpretation, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

JR: Design of the study, suggestions for data interpretation, suggestions for graphical representation of figures, and critical reviewing of the manuscript for important intellectual content.

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CHAPTER 2

A SELECTION METHOD FOR A REPRESENTATIVE TRIAL

Automatic Selection of a Representative Trial from Multiple Measurements using Principle Component Analysis

Katrin Schweizer, Philippe C Cattin, Reinald Brunner, Bert Müller, Cora Huber & Jacqueline Romkes

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ABSTRACT

Experimental data in human movement science commonly consist of repeated measurements under comparable conditions. One may face the question of how to identify a single trial, a set of trials, or erroneous trials from the entire dataset. This study presents and evaluates a Selection Method for a Representative Trial (SMaRT) based on a Principal Component Analysis. SMaRT was tested on 1841 datasets containing 11 joint angle curves of gait analysis. The automatically detected characteristic trials were compared with the choice of three independent experts. SMaRT required 1.4 s to analyse 100 datasets consisting of 8 ± 3 trials each. The robustness against outliers reached 98.8% (standard visual control). We conclude that SMaRT is a powerful tool to determine a representative, uncontaminated trial in movement analysis datasets with multiple parameters.

INTRODUCTION

Experimental data in human movement science commonly consist of repeated measurements under comparable conditions. A trial often comprises of several parameters as a function of time, such as joint angle curves. Here, the question arises on how to identify a number of characteristic trials or how to exclude erroneous trials. For simplified interpretation, the experimental data might be reduced to a single characteristic trial or to a mean of several trials to alleviate assimilation [1]. Calculating the mean, however, can filter out peaks and time shifts [2]. Regardless if one prefers to progress with one or with a mean of several trials, a defined number of uncontaminated trials from the entire dataset has to be selected.

In the literature some alternative methods to identify representative trials were proposed [3-6]. The most common approach is visual inspection [6]. While outliers and contaminated data are easily identified, the constraints of this approach lie in time consumption and lack of objectivity. Random selection of trials is fast [3], but only meaningful for entirely uncontaminated data. Duhamel et al. [4] published an algorithm to select the subset of four knee flexion/extension curves based on the intra-class correlation coefficient. Although this approach can be extended to several joint angles, it is unlikely that the same trial for each curve will be selected. The drawback of the proposal to detect one representative trial across several inter-segment angles from Carson et al. [5] is the averaging, as waveform information is neglected. Therefore, it is desirable to reveal a method, which allows to (1) identify representative trials across several joint angles, (2) be automatic and fast, (3) be reliable and avoid the subjectivity of visual inspection, and (4) to be robust against erroneous data, including labelling errors. The purpose of this work is to evaluate the Principle Component Analysis (PCA) [7], as an approach for the automatic detection of representative trials.

METHODS

Data acquisition and processing

To evaluate SMaRT, 1841 retrospective datasets, acquired from daily clinical practice between 1999 and 2010, were included. Data originated from patients with various gait disorders (1653 datasets) and healthy subjects (188 datasets). All participants signed written consent, as required by the responsible ethical committee.

A VICON motion capture system (Oxford, UK) with six cameras was used to track the trajectories of reflective markers which were attached to anatomical landmarks according to the Plug-in-Gait model [8]. Eleven joint angles were calculated and normalised to one gait cycle by means of 51 discrete values: pelvic tilt/obliquity/rotation, hip flexion/abduction/ rotation, knee flexion/abduction, ankle flexion/rotation, and foot progression.

SMaRT

SMaRT was implemented in MATLAB (MathWorks Inc., R2010a, Natick, USA) and was run separately for each body side. In the supplementary material we provide the SMaRT code.

A dataset for one subject consisted of a three-dimensional matrix X_{tif} containing a patient dependent number of trials t (3 to 18), data points i = 51, and angles f = 11.

Firstly, SMaRT applied a PCA [7] on X_{ti}, i.e. on each trial and each angle of one individual subject, separately. The output delivered PC-scores Z_{tn}, where n is the number of PC-scores, i.e. number of trials minus one. Secondly, the median M_n of the PC-scores was determined across all trials of one individual subject for the 11 angles, separately. Thirdly, the Euclidean distances d_t [9] of each trial of a subject to the median of the PC-score were computed (Figure 2.1).

After applying the three steps for each angle individually, the distances of each trial across all angles were summed. In this specific evaluation of SMaRT, the trial with the smallest overall distance to the median (Figure 2.2) was selected and defined as a representative trial.

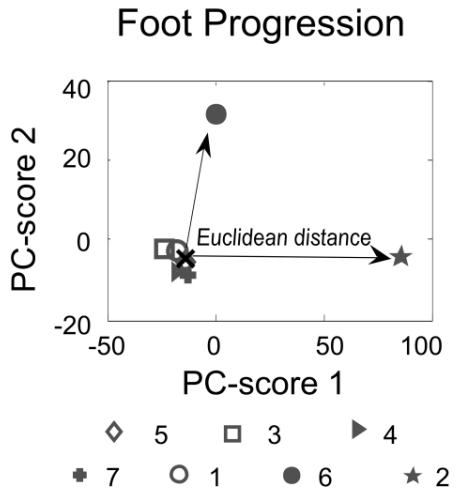

Figure 2.1: PC-scores and median for all seven trials of a single subject
The scatter plot represents the first two PC-scores for the foot progression angle during walking for each trial (symbols) of a subject. The calculated median of the PC-scores is represented with a cross.

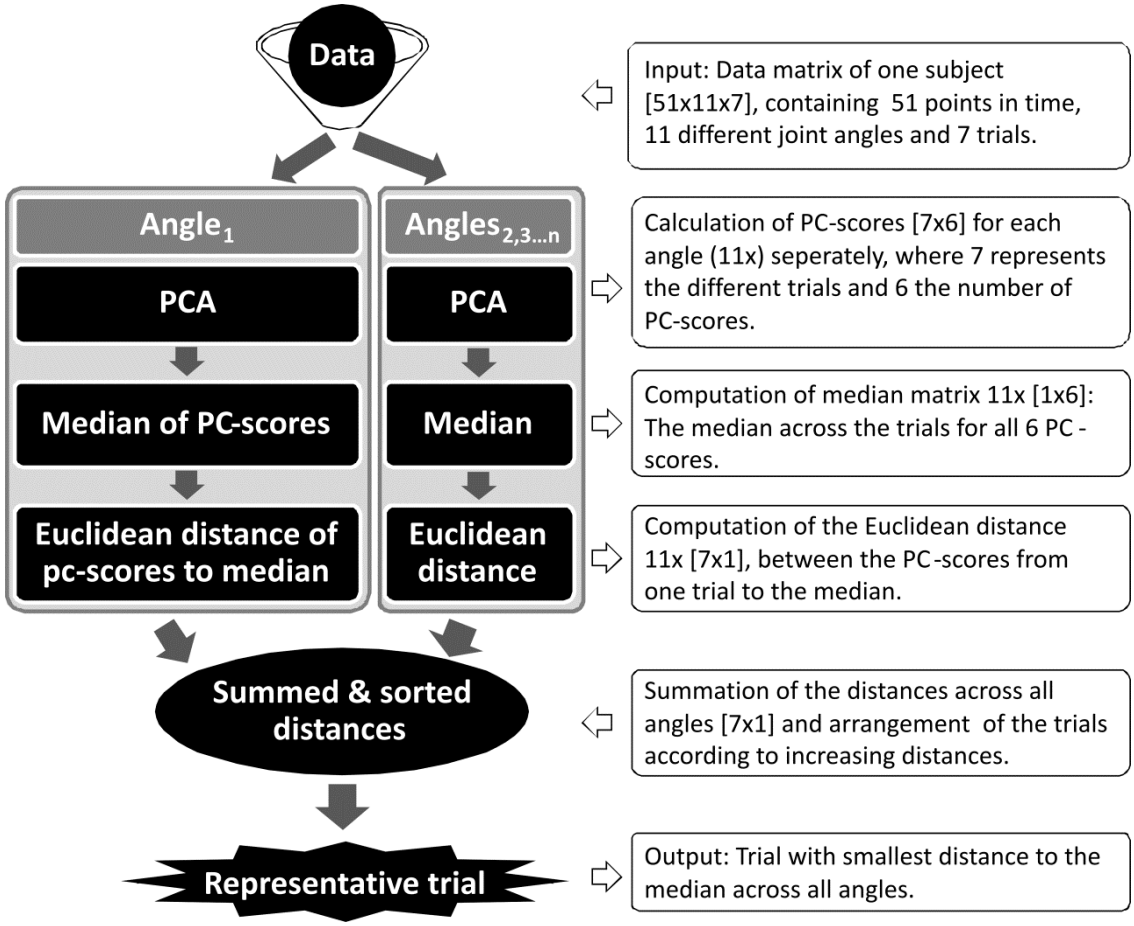

Figure 2.2: Flowchart of SMaRT, showing the single steps to select the representative trial
The black fields represent each step of the algorithm. The text boxes on the right give an accurate description of the input and output data of the single steps. As an example, we used here 11 joint angles normalised to 51 data points for 7 trials.

Evaluation of SMaRT

Two evaluation procedures were accomplished. Firstly, the robustness of SMaRT against outliers was determined by counting the false positives. The first author estimated the error rate via visual inspection of 1841 datasets.

Secondly, three experts in clinical gait analysis visually selected representative trials to be compared with the SMaRT choice. From the experimental data, 30 sets with 219 trials were randomly selected using a MATLAB routine. The experts independently worked through these datasets, where each dataset was plotted into consistency graphs containing all recorded trials for a subject. The experts assessed each trial and angle. They then decided whether the trial was representative or not. Multiple selections were allowed.

The number of representative trials on which one, two, or all three experts agreed on was expressed in percentage of the total number of trials. Additionally, the percentage of conformity between the selections of SMaRT and experts were evaluated.

RESULTS AND DISCUSSION

SMaRT, since based on PCA, is sensitive to waveforms [10]. Whereas trials with large waveform deviation (e.g. mirrored curves due to labelling errors) will have a large Euclidean distance to the median, trials with similar waveforms but with an offset from the median have a small distance. This is beneficial, as we usually consider larger waveform deviations more likely to result from measurement errors than offset curves with characteristic waveforms. Note that SMaRT does not evaluate the variability of the data. The consistency of the data could be determined by one of the methods proposed by Chau et al. [1] before running SMaRT.

Performance of SMaRT

SMaRT took 1.4 s to analyse 100 datasets consisting of 8 ± 3 trials each on a 64-bit computer (HP Compaq 8100 Elite). The three experts needed 15, 28, and 43 minutes to assess the 30 datasets. Hence, SMaRT evaluates the data, without subjective bias, more than three orders of magnitude faster than the experts. While visual and random selection can produce different results in multiple assessments, SMaRT provides full repeatability.

Evaluation of SMaRT

In datasets with contaminated trials, SMaRT selected a trial without visible sign of contamination (Figure 2.3). The first author revealed an error rate of SMaRT of 1.2%. SMaRT filters adequately erroneous data because the median, which is robust against outliers, is calculated. Hence, SMaRT operates as quality assurance where visual control is impossible due to large amounts of data. This procedure is limited to data with less than half of the trials contaminated.

The SMaRT selection agreed with those of at least one expert to 96.7% (29/30), with those of at least two experts to 80.0% (24/30), and with those of all three experts to 56.7% (17/30). SMaRT once selected a trial not chosen by the experts. This trial, not a distinct outlier, showed a small irregularity in one angle. Although selection of multiple representative trials was allowed, the inter-rater reliability between the three experts was low. The three experts agreed on 25.1% (55/219) of representative trials only, and at least two experts agreed on 44.3% (97/219). This affects the agreement between SMaRT and the choice of two, or even three experts. Nonetheless, the agreement between two experts and SMaRT is still regarded as high.

In conclusion, SMaRT meets our requirements for an objective, fast, reliable, and automatic selection tool of a characteristic trial from multiple trials containing numerous angles. Hence, the selected trial is the same trial for all curves. Additionally, the method can be used as a filter for contaminated data or as a quality assurance procedure, as it is robust against a limited number of outliers. The algorithm can either be extended to an arbitrary choice of trials or to an individually required number of parameters (e.g. kinetic parameters) or both. The suc-

cessful application of SMaRT may be profitably applicable to any kind of time series derived from movement analysis.

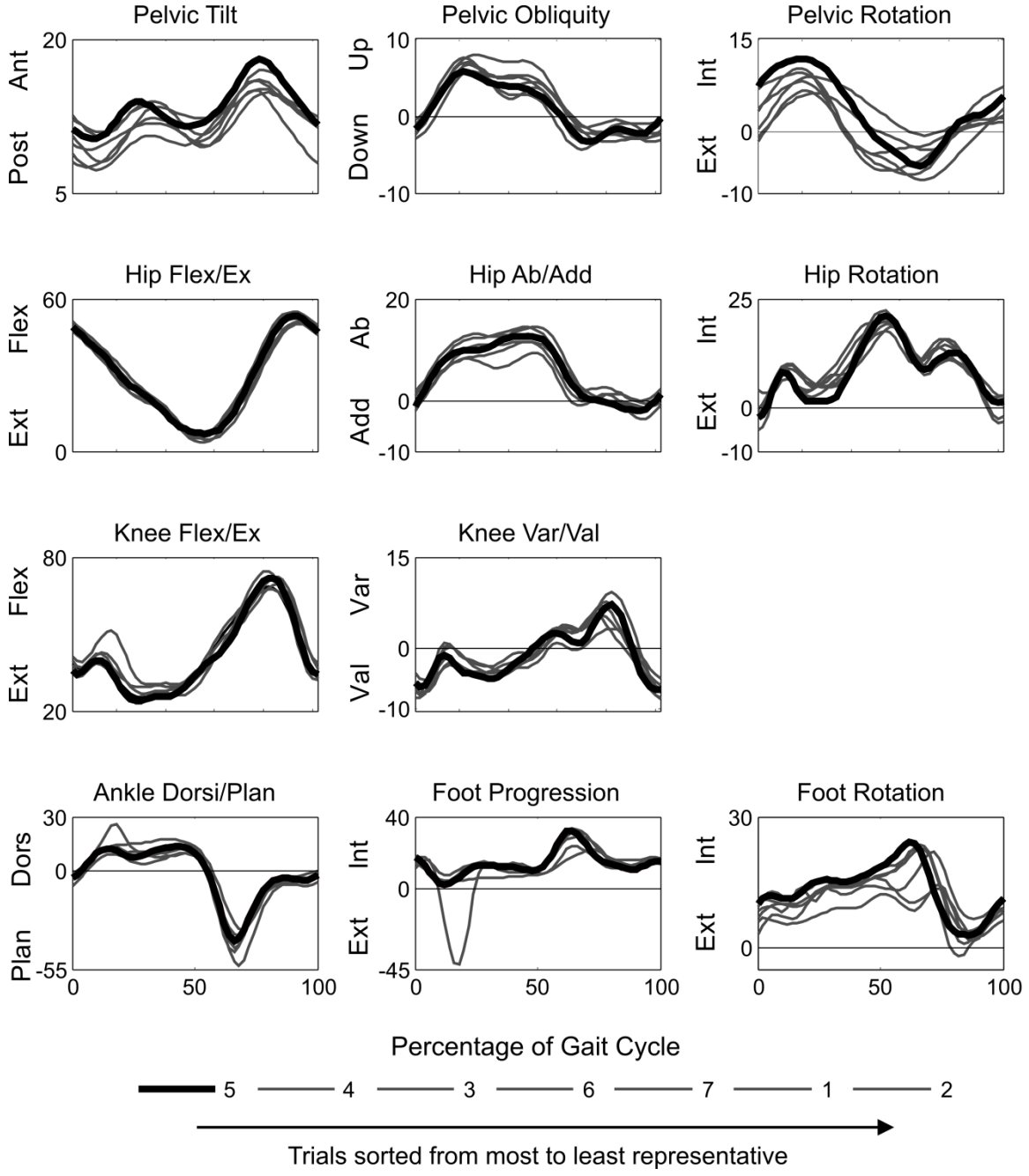

Figure 2.3: Consistency plot of all seven trials for one subject
The joint angle curves are plotted for all seven trials of one dataset, representing a single subject. The representative trial (bold line) is the trial selected by the algorithm.

ACKNOWLEDGEMENT

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Further the authors appreciate the work of their gait laboratory's team in data acquisition, and thank the three experts for the evaluation. We thank Corina Nüesch for providing data to check SMaRT on data normalised to 101 data points.

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CHAPTER 3

THE INFLUENCE OF MUSCLE STRENGTH ON GAIT KINEMATICS

The Influence of Muscle Strength on the Gait Profile Score (GPS) across Different Patients

Katrin Schweizer, Jacqueline Romkes, Michael Coslovsky & Reinald Brunner

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ABSTRACT

Background: Muscle strength greatly influences gait kinematics. The question was whether this association is similar in different diseases.

Methods: Data from instrumented gait analysis of 716 patients were retrospectively assessed. The effect of muscle strength on gait deviations, namely the Gait Profile Score (GPS) was evaluated by means of generalized least square models. This was executed for seven different patient groups. The groups were formed according to the type of disease: orthopaedic/neurologic, uni-/bilateral affection, and flaccid/spastic muscles.

Results: Muscle strength had a negative effect on GPS values, which did not significantly differ amongst the different patient groups. However, an offset of the GPS regression line was found, which was mostly dependent on the basic disease. Surprisingly, spastic patients, who have reduced strength and additionally spasticity in clinical examination, and flaccid neurologic patients showed the same offset. Patients with additional lack of trunk control (Tetraplegia) showed the largest offset.

Conclusion: Gait kinematics grossly depend on muscle strength. This was seen in patients with very different pathologies. Nevertheless, optimal correction of biomechanics and muscle strength may still not lead to a normal gait, especially in that of neurologic patients. The basic disease itself has an additional effect on gait deviations expressed as a GPS-offset of the regression line.

INTRODUCTION

Instrumented gait analysis provides detailed information on the gait kinematics of a tested individual under standardised laboratory conditions. Inter-individual comparison of this data can become difficult, especially if large numbers of individuals are involved. Several gait scores have been developed for the purpose of an easier general overview: the Gillette Gait Index (GGI), earlier described as the Normalcy Index [1], the Gait Deviation Index (GDI) [2], and the Gait Profile Score (GPS) [3]. These indices summarise kinematic data as a representation of the overall gait deviation as a single value. The more this index deviates from normal, the more the patient's gait is pathological. The strong point of these indices is that they can give a statistical overview over a large cohort. A weak point is that they neither provide the direction of gait deviation (e.g. below or above the norm) nor the factors contributing to the change of function. Another weak point is that they do not show whether the deviation is due to time-shifts, or if the joint curves deviate in magnitude only. Until now, such indices have been mainly used for patients with neurologic diseases. As an example, Schwartz et al. [2] found the GDI to decline with the severity of cerebral palsy when they compared the overall gait pathology in hemiplegia, diplegia, triplegia, and quadriplegia. This study revealed a coherence of the biomechanical deviation during gait with the variable geographical expression of a single basic disease, in this case the lesion of the central nervous system.

While GPS and GDI are computed on the entire joint curve, the GGI is computed on specific parameters of each curve. Therefore, it reduces the information given by each curve beforehand. The GPS was chosen for the present study as it is the most compound and neutral score in respect of the contributing parameters. It has the advantage over the GDI in that one can split the GPS up to the single joint levels, namely the GVS [3] for further analysis.

Clinical testing, such as functional muscle strength testing, delivers further valuable information to interpret gait analysis data. In the literature, muscle strength was identified as a major factor that influences gait [4-7]. However, it is unknown to date, how the widely used GPS is associated with muscle strength. Further, one does not know if muscle weakness has the same effect on the gait deviations in patients with different pathologies. Although Schwartz et al. [2] found higher gait deviations in more severe impaired cerebral palsy patients, it is necessary to investigate whether these findings were due to the increasing weakness of these patients or independent of their muscle strength.

The purpose of the present study was to investigate the association between the muscle strength of patients and the kinematic gait deviation across various pathologies. We hypothesised a negative correlation between the GPS, as a measure of the gait deviation, and the mean manual muscle strength of the leg muscles. The question posed was whether this correlation was similar across different patient groups. Additionally, we hypothesised that the severity of the pathology was reflected in a higher gait deviation in patients with normal muscle strength. Knowledge on the association between MMS and GPS in different patient groups is of high relevance as both parameters are widely used in the field of clinical gait analysis.

METHODS

In this retrospective study all three dimensional gait analysis datasets from daily clinical practice in our Laboratory for Movement Analysis were considered. Consecutive data from 2001 till 2012 were available, covering patients with different orthopaedic and neurologic pathologies. All patients signed an informed consent. The study was approved by the local ethical committee.

Subjects

Patients were included in this study when providing at least three lower body kinematic trials. Only patients walking barefoot without any assistive devices were evaluated. Subsequently, data on manual muscle strength testing [8] had to be available. In total, 716 out of 1144 patients with 46 different primary pathologies met the selection criteria. Pathology groups of comparable size were formed in order to gain overview. The groups were clustered according to the source of the problem (only orthopaedic, neurologic spastic with trunk control, neurologic spastic without trunk control, neurologic flaccid) and whether the problem was uni- or bilateral. Accordingly, seven groups were formed:

1) Orthopaedic unilateral (OUni): All problems of foot, knee, hip including true diseases such as Morbus Perthes disease, as well as simple pain, and unilateral torsional malalignment;
2) Orthopaedic bilateral (OBi): Spinal disorders without any neurologic involvement, Arthrogryposis Multiplex Congenita, leg length discrepancy, bilateral torsional malalignment;
3) Neurologic flaccid unilateral (NflaUni): Poliomyelitis, palsy of single nerves;
4) Neurologic flaccid bilateral (NflaBi): Spina bifida, paraplegia, muscle dystrophy, bilateral poliomyelitis, developmental retardation, trisomias with ligamentous laxity and muscle hypotonia;
5) Neurologic spastic unilateral (NspUni): Hemiparesis of various aetiologies;
6) Neurologic spastic bilateral with adequate trunk control (NspBi): Diplegia
7) Neurologic spastic bilateral without adequate trunk control (NspBiNTC): Tetraplegia of various aetiologies (cerebral palsy, brain injury, syndromes).

For an exact composition of the patient groups, please refer to the appendix Table S1.

Data Collection

Kinematic gait analysis data were collected by a VICON motion analysis system (six-camera system 370, 60 Hz, marker diameter 25 mm, years 2001-2002; six-camera system 460, 120 Hz, marker diameter 14 mm, years 2003-2010; twelve-camera system MXT20, 200 Hz, since 2011). Patients and controls walked on a 10 m level ground walkway at a self-selected speed.

According to the protocol of Kadaba et al. [9], fifteen passive reflective markers were fixed bilaterally to specific anatomical landmarks on the subject's legs and pelvis. For appropriate anthropometric scaling height, weight, leg length, width of ankles and knees, and tibial torsion were measured. The knee alignment device was used in the static trial to establish the knee flexion axis (Motion Lab Systems, Inc., Los Angeles, USA).

Additionally, manual muscle strength was tested by a physiotherapist (scale 0 = paralysed muscle to 5 = strong/normal) [8] prior to the gait analysis. The muscle groups accessed were hip flexors/extensors/abductors and in-/external rotators, knee flex-/extensors, plantar-/ dorsiflexors.

Data Processing and Analysis

Kinematic data were normalised to a gait cycle containing 51 data points (0-100%) using MATLAB software (MathWorks, Inc. Version R2010a, Natick, MA, USA). For each patient the GPS [3] was calculated as a quantity of overall gait deviation. For patients in groups with similar GPS, the GDI [2], GGI [1], and Gait Variable Scores (GVS) [3] were additionally calculated to distinguish gait deviations in each joint of the lower body and each body plane. The Geers' Metric, as summarised in Lund et al. [10], helped identify whether the joint angle deviated in magnitude or showed a time/phase shift. For calculation of the gait indices, 102 datasets of healthy subjects acquired in our laboratory were used. The mean manual muscle strength (MMS) of a patient was calculated by averaging all values derived by the manual muscle testing on the leg.

Primary variables of interest were the GPS and MMS. For patient groups with similar GPS, the GGI, GVS, and Geers' Metric values in sagittal plane of the pelvis, hip, knee, and ankle were compared. In frontal plane the pelvis and hip were of interest and in transversal plane the pelvic, hip, ankle, and the foot progression angle.

Statistical Analysis

One representative gait trial for each patient was automatically selected for further analysis. The selected trial was the trial closest to the median of the principal component score across all angles [11]. In unilateral impaired patients, the involved leg was investigated. For those with bilateral impairments, one leg was selected randomly.

Statistical analysis was performed with R2.12.0 [12]. To assess whether the effects of MMS level on GPS differ among pathology groups we included MMS and its interaction with patient group into the model. The results were adjusted for Body Mass Index (BMI), age, age^2, and sex. The interactions of patient group with age and age^2 were also tested. Due to heterogeneity of the data, generalised least squares were used [13]. Models with different variance structures were compared using Akaike's Information Criteria to determine the optimal variance structure. The variance structure giving the best fit, allowed for different variances per treatment group (function varIdent, R package nlme). For ease of interpretation, GPS levels between the groups were compared at a MMS of 5 (normal muscle strength), and age was

centred on its mean (17.5 years). Interactions were removed from the model when not significant ($p>0.05$). Three data points were identified as outliers and removed from the analysis, making no qualitative difference in the results but a better estimation of coefficients. Values are represented as estimates and standard errors (SE) unless otherwise specified.

Kruskal-Wallis multiple comparison testing and Mann-Whitney *U* post hoc tests with the Bonferroni-Holm adjustment were conducted to derive differences between the patient characteristics of the OUni group and the remaining patient groups. Further, Mann-Whitney *U*-tests were conducted for GVS and Geers' Metric values for groups with similar GPS, as the majority of the data was not normally distributed according to the Shapiro-Wilk test.

RESULTS

Table 3.1 describes the characteristics of the seven patient groups and healthy controls.

Table 3.1: Subject groups characteristics.
For each patient group the number of subjects (N), the sex (female/male), as well as the mean (± one standard deviation) age in years, body mass index (BMI), cadence, walking speed and step length are reported. The last three gait parameters are reported as non-dimensional parameters. The abbreviation for the patient groups are orthopaedic uni-/bilateral (OUni/OBi), neurologic flaccid uni-/bilateral (NflaUni/NflaBi), neurologic spastic uni-/bilateral with/without adequate trunk control (NspUni/NspBi/NspBiNTC). Significant differences compared to the reference group (OUni) are highlighted in bold.

Subject group	N	Sex [f/m]	Age [years] (SD)	BMI [kg/m^2] (SD)	Normalised Gait Parameters		
					Walking speed (SD)	Cadence (SD)	Step length (SD)
Controls	102	51/51	25.1 (12.0)	21.7 (3.4)	0.45 (0.05)	35.42 (2.05)	0.77 (0.07)
OUni (ref.)	93	48/45	20.9 (13.7)	21.8 (4.4)	0.43 (0.07)	34.76 (2.93)	0.75 (0.09)
OBi	176	81/95	**15.7 (8.7)**	20.5 (4.0)	0.44 (0.06)	35.12 (2.77)	0.76 (0.09)
NflaUni	12	4/8	21.8 (16.3)	19.6 (3.5)	0.41 (0.09)	33.09 (4.58)	0.76 (0.10)
NflaBi	83	41/42	19.4 (12.9)	21.5 (5.6)	**0.36 (0.09)**	**32.03 (4.45)**	**0.66 (0.13)**
NspUni	176	80/96	16.7 (10.0)	20.8 (5.1)	**0.41 (0.08)**	**33.31 (3.98)**	0.72 (0.10)
NspBi	119	46/73	15.8 (7.9)	20.0 (3.9)	**0.37 (0.09)**	**33.36 (4.33)**	**0.67 (0.12)**
NspBiNTC	57	20/37	19.1 (9.5)	20.3 (4.5)	**0.34 (0.12)**	**32.01 (6.60)**	**0.61 (0.16)**

MMS had a strong and negative effect on the GPS score (MMS: -2.9 SE 0.22, t_{701} = -13.7, p < .001). No significant differences in this relationship existed between the patient groups, as the interaction between MMS and patient group was not significant ($F_{6,695}$ = 0.5, p = .807) (Figure 3.1). However, Patient groups strongly differed in the GPS offset ($F_{6,701}$ = 6.7, p < .001) at a MMS of 5 (Table 3.2).

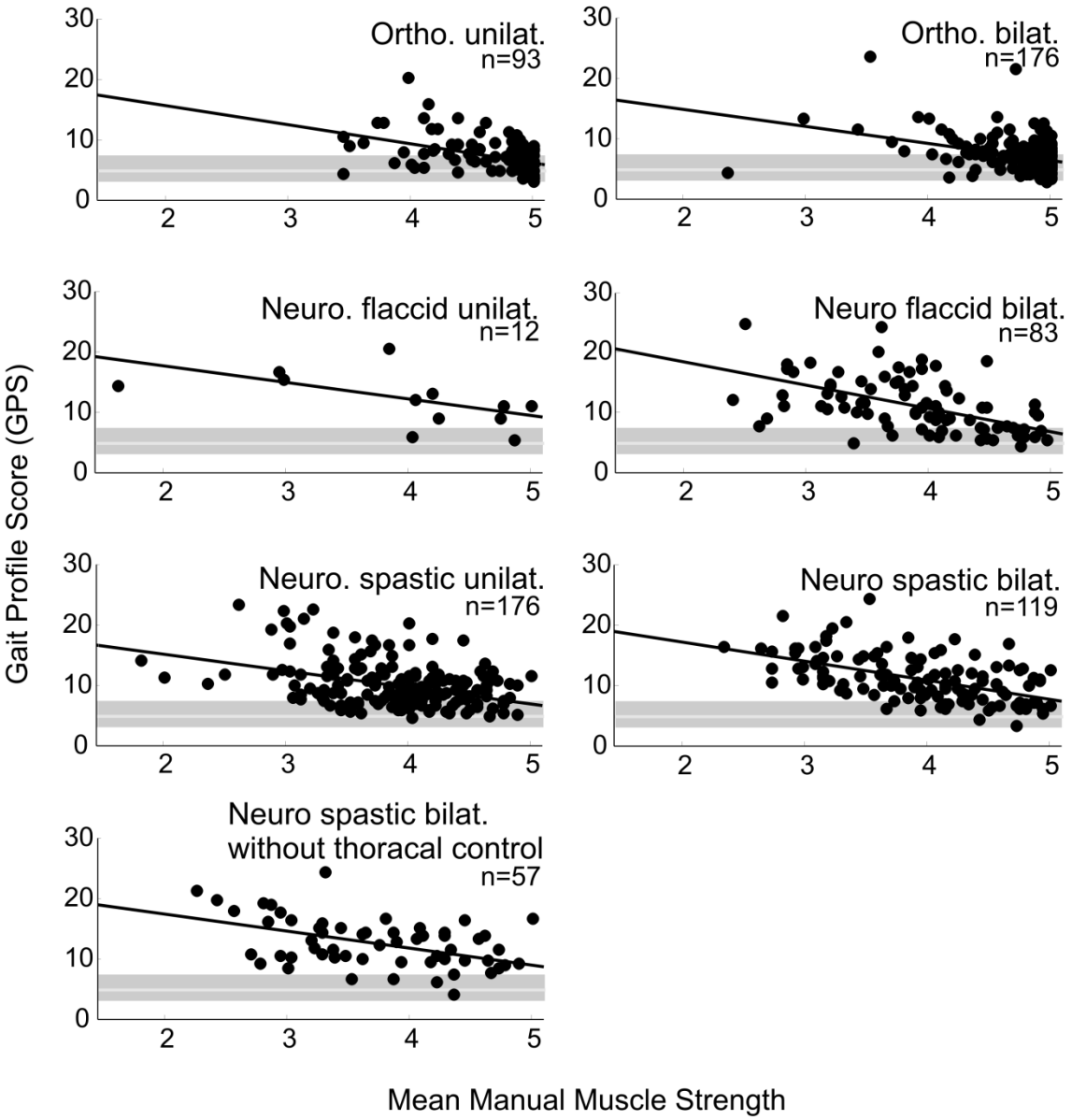

Figure 3.1: Effect of muscle strength on GPS
Regression lines and scatter plots of mean manual muscle strength (MMS) are plotted against Gait Profile Score (GPS) in the different patient groups. The grey band represents 95-GPS-percentile of the norm, and the white line marks the median of the norm. The patient groups are orthopaedic uni-/ bilateral (OUni/ OBi), neurologic flaccid uni-/ bilateral (NflaUni/ NflaBi), neurologic spastic uni-/ bilateral with/ without adequate trunk control (NspUni/ NspBi/ NspBiNTC).

Table 3.2: Summary of differences in mean Gait Profile Score (GPS) by patient groups at mean manual muscle strength (MMS) of 5.
The differences in the intercepts (Gait Profile Scores value) at the mean age (17.5 years) and at normal muscle strength (MMS = 5) of each patient were compared to the reference group OUni. Hence, the sum of the intercept of OUni and another patient group estimates the GPS of this group. Coefficients are expressed per year of age relative to the centred age. Interaction values represent the differences in the strength (slope) of the effect of centred age (AgeC = 17.5 years) in the different groups. The abbreviation for the patient groups are orthopaedic uni-/bilateral (OUni/OBi), neurologic flaccid uni-/bilateral (NflaUni/NflaBi), neurologic spastic uni-/bilateral with/without adequate trunk control (NspUni/NspBi/NspBiNTC). Standard error (SE) of the intercepts, *t*-values and significance of the difference (*p*) are reported. Significant differences are highlighted in bold.

Patient group	Intercept (SE)	*t*	*p*-value
OUni (reference)	4.9 (0.7)	------	------
OBi	0.1 (0.3)	0.34	.739
NspUni	0.5 (0.4)	1.24	.217
NspBi	**1.7 (0.5)**	**3.83**	**<.001**
NflaBi	**1.7 (0.5)**	**3.22**	**.001**
NspBiNTC	**2.5 (0.6)**	**4.08**	**<.001**
NflaUni	**3.1 (1.0)**	**3.07**	**.002**
AgeC:OUni (reference)	**-0.09 (0.04)**	**-2.25**	**.025**
AgeC:OBi	-0.04 (0.03)	-1.31	.190
AgeC:NspUni	0.02 (0.03)	0.65	.511
AgeC:NspBi	0.04 (0.04)	0.86	.391
AgeC:NflaBi	**-0.08 (0.04)**	**-2.21**	**.028**
AgeC:NspBiNTC	0.08 (0.06)	1.41	.158
AgeC:NflaUni	**-0.15 (0.06)**	**-2.38**	**.018**

In comparison, our healthy controls had a median GPS of 4.8° (interquartile range 3.9-5.8). The GPS offset of NspUni compared to the reference group (OUni) was minimal (0.5) and similar to OBi (0.1). It was larger with NspBiNTC (2.5) and NflaUni (3.1). The offsets of NflaBi and NspBi were similar (1.7). Figure 3.2 displays the mean angles of the NspBi and NflaBi. Table 3.3 lists the differences between these two groups concerning the single joint levels and body planes in GVS and Geers' Metric.

The interaction patient group*age^2 was not significant, and was removed from the model ($F_{6,681}$ = 0.792, p = 0.577). The effect of age on GPS was quadratic, and depended on patient gender (age*sex: $F_{1,701}$ = 9.5, p = .002; age^2*sex: $F_{1,701}$ = 5.9, p = .015). The marginal effects of age and sex on GPS can be summed as GPS = -0.09 x age + 0.002 x age^2 for females and GPS = 0.16 + 0.03 x age + 0.006 x age^2 for males.

BMI had a positive effect on GPS score (0.08 SE 0.03, $F_{1,701}$ = 6.5, p = .011). Age had a significantly different effect on GPS score amongst patient groups ($F_{1,701}$ = 3.1, p = 0.005). Table 3.2 lists the slope of the age effect for each group separately.

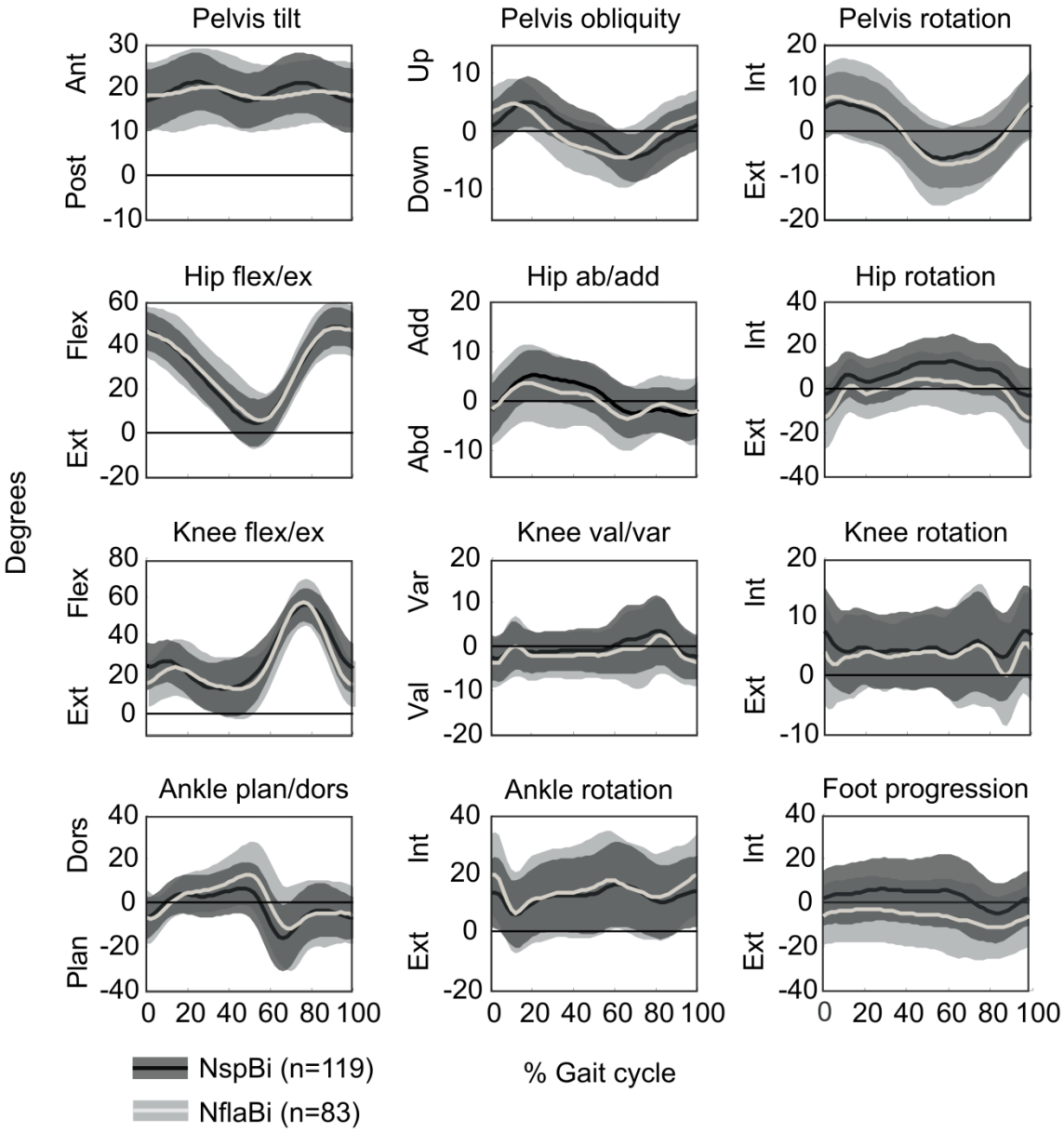

Figure 3.2: Joint angles of patients with spasticity and flaccid muscles.
Mean joint angles with ± one standard deviation band of patients with spasticity bilateral (NspBi = black) and patients with neurologic involvement and flaccid muscles (NflaBi = gray). The data are time normalised to 0-100% of a gait cycle (x-axis) and expressed in degrees (y-axis).

Table 3.3: Differences between spastic patients and patients with flaccid muscles in various gait indexes.

The median of each group and the interquartile ranges are stated, as well as statistical differences between the two groups (*p*-values). Significant differences are highlighted in bold. Rows denote the different gait indexes. List of abbreviations: NspBi = patients with neurologic impairment and spasticity bilateral, NflaBi = neurologic flaccid bilateral patients, GDI = Gait Deviation Index, GGI = Gilette Gait Index, GPS = Gait Profile Score, GVS = Gait Variable Score, Geers = Geers Metric where M = magnitude and P = phase shift, flex/ex = flexion/extension, plan/dors = plantar/dorsiflexion, ab/add = ab-/adduction, rot = rotation.

	NspBi		NflaBi		
Index	Median	Range	Median	Range	*p*-value
GDI kinematic	75.3	(66.4-83.8)	75.8	(65.6-86.3)	.868
GGI	157.7	(71.6-175.4)	144.6	(57.7-156.7)	.143
GPS	11.3°	(8.3°-14.1°)	11.5°	(7.5°-14.5°)	.907
GVS pelvic flex/ex	10.0°	(5.4°-13.8°)	9.8°	(4.7°-13.0°)	.501
GVS hip flex/ex	11.5°	(6.8°-15.1°)	13.3°	(7.5°-18.3°)	.071
GVS knee flex/ex	14.0°	(8.9°-17.3°)	12.8°	(7.8°-16.8°)	.172
GVS ankle plan/dors	**10.0°**	**(5.7°-11.3°)**	**12.6°**	**(6.8°-17.1°)**	**.002**
GVS pelvic ab/add	3.8°	(2.2°-5.1°)	3.9°	(2.4°-4.4°)	.878
GVS hip ab/add	5.0°	(3.1°-6.1°)	5.9°	(3.3°-7.5°)	.255
GVS pelvic rot	6.6°	(4.0°-8.2°)	7.0°	(3.4°-8.2°)	.461
GVS hip rot	12.2°	(6.7°-16.9°)	11.3°	(5.8°-13.8°)	.216
GVS foot rot	14.1°	(7.3°-19.5°)	13.4°	(6.4°-18.1°)	.457
Geers pelvic flex/ex (M)	90.9	(47.1-131.7)	86.0	(38.2-128.4)	.528
Geers pelvic flex/ex (P)	**4.9**	**(2.8-6.9)**	**4.3**	**(2.1-5.8)**	**.010**
Geers pelvic ab/add (M)	79.2	(28.2-107.0)	84.2	(14.6-125.7)	.642
Geers pelvic ab/add (P)	26.7	(17.1-35.5)	27.9	(16.4-38.3)	.695
Geers pelvic rot (M)	174.6	(93.4-240.7)	201.1	(63.1-256.7)	.876
Geers pelvic rot (P)	27.5	(18.3-35.9)	25.8	(17.1-32.8)	.509
Geers hip flex/ex (M)	27.8	(11.8-43.7)	31.4	(1.2-58.1)	.903
Geers hip flex/ex (P)	**7.9**	**(5.3-10.2)**	**9.9**	**(6.6-13.0)**	**.002**
Geers hip ab/add (M)	48.7	(9.9-77.2)	74.6	(19.6-94.3)	.071
Geers hip ab/add (P)	33.1	(23.4-42.8)	34.8	(22.0-47.3)	.644
Geers hip rot (M)	155.2	(50.9-229.0)	141.6	(51.9-184.0)	.386
Geers hip rot (P)	34.4	(28.2-39.4)	34.3	(25.7-41.7)	.667
Geers knee flex/ex (M)	**6.9**	**(-9.7-17.9)**	**2.0**	**(-15.8-13.1)**	**.048**
Geers knee flex/ex (P)	10.8	(7.0-14.1)	10.6	(7.0-14.5)	.777
Geers ankle plan/dors (M)	**35.4**	**(-12.4-42.2)**	**67.9**	**(-1.0-109.9)**	**.001**
Geers ankle plan/dors (P)	28.4	(19.7-35.6)	31.2	(22.2-40.2)	.079
Geers ankle rot (M)	**99.5**	**(-6.2-187.4)**	**139.7**	**(30.9-201.4)**	**.043**
Geers ankle rot (P)	25.8	(14.3-26.9)	24.3	(13.9-24.2)	.616
Geers foot rot (M)	72.6	(-10.2-114.6)	98.5	(6.8-184.2)	.073
Geers foot rot (P)	**48.8**	**(19.4-74.2)**	**33.8**	**(13.7-51.0)**	**<.001**

DISCUSSION

In this study, the association between the gait deviation (GPS) and muscle strength (MMS) in various patient groups was investigated. The GPS is one single number which expresses the degree of gait pathology in an individual.

The results showed a clear dependence of gait deviations, assessed by the GPS, on muscle strength represented by a negative correlation. It is interesting that this correlation does not differ in the various pathology groups. Although the GPS does not represent a specific gait pattern, the severity of gait affection seems to depend on muscle strength. Even though Figure 1 yields the impression that the correlation is linear, this must be taken cautiously considering the MMS is a categorical ordered scale and not truly metric.

Our healthy controls had a similar GPS (4.8°) than the reference group in Baker et al. [3] with 5.2°. Neurologic diseases show a constant offset of the GPS for all muscle strength levels. The reference group OUni has an almost normal GPS at normal muscle strength. OBi and NspUni are only slightly higher and are still in the interquartile range of the norm. This implies that these three patient groups can still produce normal gait patterns given the fact that they have normal muscle strength. In contrast, NflaBi, NspBi, and NspBiNTC patients can hardly ever reach normal gait values. The NflaUni group GPS values may be difficult to explain, however, they could be biased due to the small group size.

It is further surprising that for well comparable groups, such as NspBi and NflaBi, the GPS offset is above the Minimal Clinically Important Difference [14] to OUni, and is equal for both groups. This is interesting as NspBi have a neurologic pathology and weakness similar to NflaBi, and one would expect spasticity to contribute to a higher gait deviation in comparison with NflaBi. However, this was not reflected in any of the gait indices GPS, GGI or GDI. This is probably due to their good correlation with each other [2,3]. The visual inspection of the joint angle curves confirmed these results, as there were no greater visual differences between the curves of NspBi and NflaBi. The significant difference in the GVS ankle plantarflexion angle between these two groups resulted mainly from the magnitude offset as discovered by the Geers' Metric. Although the Geers' Metric results disclosed further significant phase shifts at the pelvic, hip, and knee flexion angle, the absolute differences of the means were within 0.6-4.9%, which is rather low. The differences in ankle rotation magnitude and in foot progression phase shift were higher, however, these are not the most reliable and relevant angles in the model. Furthermore, some of the significant results in Table 3.3 might result from multiple testing, which was not corrected for.

Similarly, the group NspUni differed only slightly from OUni, which again did not show a clear effect of spasticity. These results raise the questions: how much does spasticity influence the gait pattern, and how important is spasticity to gait deviations at least in patients with good trunk control?

The large GPS offset of NspBiNTC implies that the lack of trunk control adds additional difficulties to walking. These patients, mainly with tetraparetic cerebral palsy, present more global stiffness than patients with a more hyperreflexic type of spasticity like diplegics. It is

impossible to separate the effect of stiffness from the lack of trunk control, but probably these two factors are linked.

GPS increase depends on muscle strength. The severity of the basic disease adds an offset which limits the best possible result of treatment. Spasticity seems to be of minor importance whereas trunk control has a major effect on gait. Other factors may change the biomechanics, which at least to some degree also depend on the basic disease. For instance, extensibility of a joint, e.g. knee hyperextension, reduces the need of muscle strength to control posture [15]. However, some diseases have an increased tendency to flexion deformity, such as cerebral palsy [16]. Thus, some basic diseases can cope better with muscle weakness than others, which are limited in their compensatory availability.

All modelled interactions only had the function to control their effect on the model. The results are not extensively discussed here. First, because they were not subject to the main research question, and second, because their effects were small and should not be overemphasised.

The study has some weaknesses. Some patient groups, such as OUni, OBi, and NflaBi, included patients with diverse diagnoses, whereas other groups, such as NspUni and NspBiNTC, were more homogenous. The groups themselves differed in the distribution of their basic affection severity and in their mean age. The orthopaedic groups included more patients with normal muscle strength than the groups with neurologic diseases.

Furthermore, the results merely apply to unassisted ambulant patients. Patients walking with assistive devices might behave differently. However, excluding these patients was inevitable, as walking aids stabilise the body, which in turn would distort the results.

Other than measuring muscle strength with a dynamometer or an isokinetic machine, the manual muscle strength testing (MMST) is not highly exact and reliable by its nature [17]. Some levels are less clearly defined than others: for instance, a value of 5 may be something between strong and extremely strong, whereas 3 is well defined as full activity against gravity. Therefore, MMST provides only a general overview on muscle strength. In literature the intra-rater reliability for MMST was estimated medium to good with a weighted Kappa between 0.71-0.93 depending on the muscle group tested [18]. Inter-rater reliability was estimated between 0.76-0.88 (intra-class correlation coefficient) for trained examiners [17]. Although more reliable tests of muscle strength exist [17], MMST is a widely used examination in daily clinical practise [18,19]. The advantages of its quick execution and the applicability to different patients, especially children, often overcome its weaknesses in clinical evaluation [19]. In order to ensure the highest reliability possible, our physiotherapists participate in yearly trainings.

This study found a negative correlation of muscle strength with the gait deviation. Besides muscle strength, the basic disease also has a direct effect on gait deviations. This was represented by the offset of the data which was independent of muscle strength. This aspect is of great interest as it may explain why, in spite of therapeutic interventions and surgical corrections, neurologic patients hardly ever reach normal gait values. The remaining offset could be caused by the neurologic disease which remains after the correction of gait biomechanics.

However, it is interesting that spasticity did not increase the offset further than weakness, at least in cases with good trunk control. This may indicate that spasticity contributes much less to gait deviations as commonly expected, and may be overestimated in daily clinical practise.

CONCLUSION

In conclusion, gait kinematics depend on muscle strength. This correlation is independent of the basic disease. The basic disease, however, adds a constant factor which depends on the severity of the basic affection. Spasticity seems to play only a minor role in gait deviations as long as trunk control is adequate, whereas muscle strength and neurologic impairment have a major impact.

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CHAPTER 4

THE INFLUENCE OF MUSCLE STRENGTH AND EQUINUS GAIT ON EMG

The Association between Premature Plantarflexor Muscle Activity, Muscle Strength, and Equinus Gait in Patients with Various Pathologies

Katrin Schweizer, Jacqueline Romkes & Reinald Brunner

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ABSTRACT

This study provides an overview on the association between premature plantarflexor muscle activity (PPF), muscle strength, and equinus gait in patients with various pathologies. The purpose was to evaluate whether muscular weakness and biomechanical alterations are aetiological factors for PPF during walking, independent of the underlying pathology. In a retrospective design, 716 patients from our clinical database with 46 different pathologies (orthopaedic and neurologic) were evaluated. Gait analysis data of the patients included kinematics, kinetics, electromyographic activity (EMG) data, and manual muscle strength testing. All patients were clustered three times. First, patients were grouped according to their primary pathology. Second, all patients were again clustered, this time according to their impaired joints. Third, groups of patients with normal EMG or PPF, and equinus or normal foot contact were formed to evaluate the association between PPF and equinus gait. The patient groups derived by the first two cluster methods were further subdivided into patients with normal or reduced muscle strength. Additionally, the *phi* correlation coefficient was calculated between PPF and equinus gait. Independent of the clustering, PPF was present in all patient groups. Weak patients revealed PPF more frequently. The correlations of PPF and equinus gait were lower than expected, due to patients with normal EMG during loading response and equinus. These patients, however, showed higher gastrocnemius activity prior to foot strike together with lower peak tibialis anterior muscle activity in loading response. Patients with PPF and a normal foot contact could possibly be applying the plantarflexion-knee extension couple during loading response. While increased gastrocnemius activity around foot strike seems essential for equinus gait, premature gastrocnemius activity does not necessarily produce an equinus gait. We conclude that premature gastrocnemius activity is strongly associated with muscle weakness. It helps to control the knee joint under load independent from the underlying disease, and it is therefore a secondary deviation. If it is treated as a primary target, then caution should be exercised.

INTRODUCTION

Three dimensional (3D) gait analysis is applied to prescribe treatment interventions in patients with different pathologies. The range spans from patients with orthopaedic impairments to flaccid muscles as well as patients with spasticity [1]. Due to anatomical and functional restrictions, patients typically reveal gait deviations, such as premature plantarflexor muscle activity (PPF) during the loading response of walking [2-8].

In the literature, abnormal plantarflexor timing is mainly described in association with initial forefoot contact [2,3,9] and/or neurological impairment [5,7,8]. For years, PPF was thought to result from spasticity or poor neuromuscular control in neurological patients [6,10]. More recent studies, however, have claimed PPF to be a secondary deviation [4,5,9,11]. According to Schmid et al. [12], secondary deviations are either passive secondary effects that follow as a physical mechanism to the primary deviation, or active compensations. The named studies came to the conclusion, that muscular weakness has been shown to provoke abnormal electromyographic (EMG) activity in orthopaedic patients with different impairments [4]. Hereby, no anatomical relationship between a specific weak muscle and a muscle showing abnormal EMG timing was possible. The medial gastrocnemius muscle was the most frequently involved muscle with abnormal EMG timing in orthopaedic patients [4]. This conforms to the findings of Goldberg et al. [13] where the plantarflexors were able to compensate for weakness in most of the major muscle groups in a forward dynamics simulation. Subsequently, PPF can result from biomechanical alteration alone, given that similar abnormal muscle activity patterns were observed in healthy subjects when mimicking the walking pattern of patients [5,9,11].

Although PPF can be observed in patients with various pathologies [3], the prevalence across different patient groups, such as in patients with neurological or orthopaedic impairments is still unknown. Muscle weakness and biomechanical alterations, seen as aetiological factors for PPF, were only examined in orthopaedic patients or healthy subjects. Therefore, it remains indistinct whether these are aetiological factors for all patients, independent of the primary pathology.

The objective of the present study was to provide an overview of the association between initial equinus foot contact, muscle strength, and PPF during walking in patients with various pathologies. We hypothesised that PPF is present in all patient groups, and that muscle weakness and equinus gait are aetiological factors for PPF independent of the pathology. The outcome is of clinical relevance, as it will assist in interpreting PPF as a primary or secondary deviation. For clinicians this distinction is crucial. Whereas a primary deviation requires treatment, secondary deviations resolve spontaneously once the primary abnormality is treated [14,15]. Consequently, the results of this study may improve treatment planning and therapy outcome.

METHODS

We retrospectively examined our 3D gait analysis database that has been collected in our laboratory for movement analysis in the context of everyday clinical practice. The parameters included for all patients were spatiotemporal parameters, lower body kinematics and kinetics, a clinical examination including manual muscle strength testing, and EMG recordings.

Subjects

All 1144 patients of the consecutive clinical gait analysis database from 2001 till 2012 were considered for this study. The database comprised of patients with various orthopaedic and neurologic pathologies, mainly children and adolescents, but also adults. Selected for this study were all patients who walked barefoot without any assistive devices. They also should have provided complete EMG, kinetic and kinematic data for at least three trials, as well as a complete manual muscle strength testing. Finally, 716 patients with 46 different primary pathologies and 102 healthy controls were included. All subjects signed an informed consent at the time of the gait analysis. The study was approved by the local ethical committee.

Patient Group Clustering

All patients were clustered three times according to different aspects. Subsequently, two subgroups for each group derived by the second and third clustering method were separated according to mean manual muscle strength (MMS) [19]. Subgroup "almost normal MMS" has an MMS equal to/above 4.5, and subgroup "reduced MMS" has an MMS of less than 4.5.

First clustering strategy: patients were grouped according to their diagnoses, referred to as pathology groups. Here, the primary source of the problem was of interest: orthopaedic impairments, neurologic spasticity with trunk control, neurologic spasticity without trunk control, and neurologic flaccid patients. Furthermore, it was distinguished whether the impairment was uni- or bilateral. Therefore, seven groups were formed that are described in Table 4.1. Please refer to the appendix for a more detailed composition of the patient groups.

Second, independently of the first clustering, the entire patient population was clustered according to the impaired joint level, namely impairment groups, to avoid bias by the second clustering. A joint was defined to be impaired if the kinematic deviation in sagittal plane was above the 97.5 percentile of the Gait Variable Score (GVS) [16] for our controls. The thresholds were for the hip 10.9°, knee 11.0°, and for the ankle 7.2°. This resulted in eight impairment groups: 1) patients with abnormal hip; 2) patients with abnormal knee; 3) patients with abnormal ankle 4) patients with abnormal hip and knee; 5) patients with abnormal hip and ankle; 6) patients with abnormal knee and ankle; 7) patients with abnormal hip, knee, and ankle; 8) patients with normal hip, knee, and ankle joints.

Table 4.1: The seven pathology groups.

Group abbreviation	Type of impairment	Muscle tone	Topographical description of the impairment	Included pathologies
OUni	Orthopaedic	-	Unilateral	All problems of foot, knee, hip, including true diseases such as Morbus Perthes, as well as solely pain, and unilateral torsional malalignment
OBi	Orthopaedic	-	Bilateral	Spinal disorders, Arthrogryposis Multiplex Congenita, leg length discrepancy, torsional malalignment
NflaUni	Neurologic	Flaccid	Unilateral	Poliomyelitis, palsy of single nerves
NflaBi	Neurologic	Flaccid	Bilateral	Spina bifida, paraplegia, muscle dystrophy, bilateral poliomyelitis, developmental retardation, trisomias
NspUni	Neurologic	Spastic	Unilateral	Hemiparesis of various aetiology
NspBi	Neurologic	Spastic	Bilateral with adequate trunk control	Diplegia
NspBiNTC	Neurologic	Spastic	Bilateral without adequate trunk control	Tetraparesis of various aetiologies

Third, again the whole patient population was divided in patients with normal EMG or PPF, and equinus or normal foot contact to evaluate the association between PPF and equinus gait.

Clinical Gait Analysis

Three dimensional gait analysis data were collected and pre-processed by a VICON motion analysis system (years 2001-2002: six-camera system 370, 60 Hz, marker diameter 25 mm; VICON Clinical Manager software; years 2003-2010: six-camera system 460, 120 Hz, marker diameter 14 mm, VICON Workstation software; since 2011: twelve-camera system MXT20, 200 Hz, VICON Nexus software; VICON, Oxford, UK). Controls and patients walked at a self-selected speed on a 10m level ground walkway. Kinetic data were acquired by two force platforms at a sampling rate of 2520 Hz (2001-2007) and of 2400 Hz since 2007 (KISTLER Instruments AG, Winterthur, Switzerland).

For the kinematics, fifteen passive reflective markers were fixed to specific anatomical landmarks bilaterally on the subject's legs and pelvis according to the protocol of Kadaba et al. [17]. Height, weight, leg length, width of ankles and knees, and tibial torsion were measured

clinically for appropriate anthropometric scaling. A knee alignment device was used for the static trial (Motion Lab Systems Inc., Los Angeles, USA).

Surface EMG was recorded simultaneously. Bipolar Ag/AgCl surface electrode pairs (10 mm diameter, 22 mm inter-electrode spacing) were placed bilaterally on the gastrocnemius medialis muscle (GM) and tibialis anterior muscle (TA) according to the SENIAM guidelines [18]. The ground electrode was placed over the tibial tuberosity. The electrodes were connected to single differential amplifiers with integrated band-pass filters at 10–700 Hz (Biovision AG, Wehrheim, Germany). The pre-amplifiers and electrodes remained the same for all measurements. Between 2001 and April 2007, pre-amplified EMG signals were collected using a Zebris System (Zebris, Tübingen, Germany) and sampled at a rate of 2520 Hz. Since May 2007 signals were collected by a Neurodata System (Neurodata, Vienna, Austria) at a sampling frequency of 2400 Hz.

Gait events, i.e. foot strike and toe-off, were set manually, and the kinematic and kinetic data were filtered by the Woltring filter (mean squared error set to 10) in the VICON software pipeline.

During the clinical examination, a physiotherapist assessed muscle strength for the lower extremity muscles of each patient according to the manual muscle strength scale described in Hislop et al. [19] (scale 0 = paralysed muscle to 5 = strong). The muscle groups accessed were: hip flexors/extensors/abductors and in-/external rotators, knee flex-/extensors, plantar-/dorsiflexors. The average on all leg muscles formed the MMS.

Data Processing

The entire post-processing and all calculations were done using the MATLAB software (MathWorks, Inc. Version R2010a, Natick, USA). Kinematic and kinetic data were normalised to a fixed amount of 51 data points per gait cycle (0-100%). A gait cycle was defined as the time between two consecutive foot strikes of the same leg. Subsequently, one trial (gait cycle) for each patient and control subject was selected using the SMaRT method [20]. Hereby, the distance of each principal component score to the median of all trials was calculated for each angle, and the trial which is closest to the median across all angles was then selected [20].

Raw EMG signals were visually inspected for artefacts and noise, before they were filtered by a 4^{th} order Butterworth band-pass filter with a cut-off frequency of 20-500Hz [21,22]. Subsequently, the signal was full-wave rectified, and a moving average was calculated with a 39.8 seconds time window similar to Romkes et al. [5]. The EMG signal was further normalised for stance (31 data points) and swing phase (20 data points), delivering together a gait cycle of 51 data points. Finally, the EMG was amplitude-normalised to the average value of each cycle.

Parameter Definition

Walking speed, cadence, and step length were evaluated in non-dimensional values according to Hof [23]. The GVS were calculated for all patients as a quantity of the kinematic gait deviation by using our own normative data.

A muscle was defined to be abnormally active if the normalised EMG signal was above a certain percentage of its peak value which was set according to the walking speed [24]. The thresholds were 28%, 23% and 31% for the non-dimensional walking speeds [23] of <0.227, 0.228-0.363, and >0.363. PPF was specified as activity of the GM above the threshold during loading response phase of gait (i.e. 0% to 10% of the gait cycle). Validity of this method was examined by checking how many of our control subjects showed "abnormal" EMG when applying this method. The less observed the better.

Equinus at initial contact was defined as 5° of plantarflexion or more at initial contact. This corresponded to approximately two standard deviations (1 SD = 2.8°) below the mean (1.2°) of the norm. Additionally, to exclude patients with a drop foot pattern, plantarflexion had to increase by at least 5 degrees over the last 10% of the gait cycle. If the latter was not fulfilled, but the ankle angle stayed 5° or more in plantarflexion during the entire gait cycle, it was still defined as equinus gait. The ankle position was termed as "normal foot contact" when the sagittal ankle angle did not meet any of the criteria for an equinus gait.

The variables and gait phases of interest were: MMS, mean GM activity during loading response and terminal swing (87-100% of the gait cycle [6]), mean TA activity during terminal swing and peak activity in loading response, equinus gait at initial contact, and mean ankle power during loading response.

Statistics

In unilateral impaired patients, the involved leg side was analysed. In those with bilateral impairments, one leg was selected randomly. Randomisation was achieved by creating a binary vector of 716 rows with the "randi" function in MATLAB.

The *phi* correlation coefficients for each pathology group were calculated between the two dichotomous variables PPF and equinus gait. As the majority of the kinetic and EMG data were not normally distributed according to the Shapiro-Wilk test, non-parametric statistics were applied to reveal significant differences. Mann-Whitney *U* tests were conducted for the GM activity of patients with normal EMG and equinus versus patients with normal ankle for the mean of loading response phase and terminal swing separately. The same patient groups were compared through Mann-Whitney *U* tests on the mean differences of their TA activity during terminal swing. They were performed as well on the peak differences in the muscle during loading response. The level of significance was set at 5% for all tests.

Prevalence of PPF within the different pathology and impairment groups is given as a percentage of the total number of patients in this group. The influence of muscle weakness on PPF was qualitatively assessed.

RESULTS

Characteristics of the control and patient groups are specified in Table 4.2. PPF was identified in 38.7% (277/716) of all patients. It was equally distributed for both genders, with 38.8% (124/320) in females and 38.6% (153/396) in males. Abnormal EMG was unevenly distributed but present in all pathology groups (Figure 4.1a). Furthermore, PPF was observed in none of the subjects within the control group.

Table 4.2: Characteristics of the subject groups.
The number of subjects (N), the mean (± one standard deviation) age in years, the sex (female/male), as well as mean (± one standard deviation) of the Body Mass Index (BMI), step length, walking speed, and cadence are reported for the healthy controls and each patient group. The last three gait parameters are reported as non-dimensional parameters [ND]. The patient groups are decoded as follows: O/N = orthopaedic/neurologic, Uni/Bi = unilateral/bilateral involvement, fla/sp = flaccid/spastic muscles, NTC = no thoracal control.

Patient group	N	Age [years]	Sex [f/m]	BMI [kg/m^2]	Step length [ND]	Walking speed [ND]	Cadence [ND]
Controls	102	25.1 (±12.0)	51/51	21.7 (±3.4)	0.77 (±0.07)	0.45 (±0.05)	35.42 (±2.05)
OUni	93	20.9 (±13.7)	48/45	21.8 (±4.4)	0.75 (±0.09)	0.43 (±0.07)	34.76 (±2.93)
OBi	176	15.7 (±8.7)	81/95	20.5 (±4.0)	0.76 (±0.09)	0.44 (±0.06)	35.12 (±2.77)
NflaUni	12	21.8 (±16.3)	4/8	19.6 (±3.5)	0.76 (±0.10)	0.41 (±0.09)	33.09 (±4.58)
NflaBi	83	19.4 (±12.9)	41/42	21.5 (±5.6)	0.66 (±0.13)	0.36 (±0.09)	32.03 (±4.45)
NspUni	176	16.7 (±10.0)	80/96	20.8 (±5.1)	0.72 (±0.10)	0.41 (±0.08)	33.31 (±3.98)
NspBi	119	15.8 (±7.9)	46/73	20.0 (±3.9)	0.67 (±0.12)	0.37 (±0.09)	33.36 (±4.33)
NspBiNTC	57	19.1 (±9.5)	20/37	20.3 (±4.5)	0.61 (±0.16)	0.34 (±0.12)	32.01 (±6.60)

All of the following results are visual trends derived by Figure 4.1. For all pathology groups, except for the NflaUni group, the percentage of patients with PPF increased from the subgroups "almost normal MMS" to "reduced MMS" (Figure 4.1a). When grouping the patients according to their impaired joints derived by the GVS, PPF was observed in all patient groups once again (Figure 4.1b). Patients with normal GVS values; hence with normal sagittal plane kinematics for all joints, have the smallest rate of incidence, followed by patients with abnormal kinematics for one of the joints. Patients with two or all three joints impaired showed the highest prevalence of PPF. Furthermore, PPF is more frequent in weak patients than in patients with normal muscle strength.

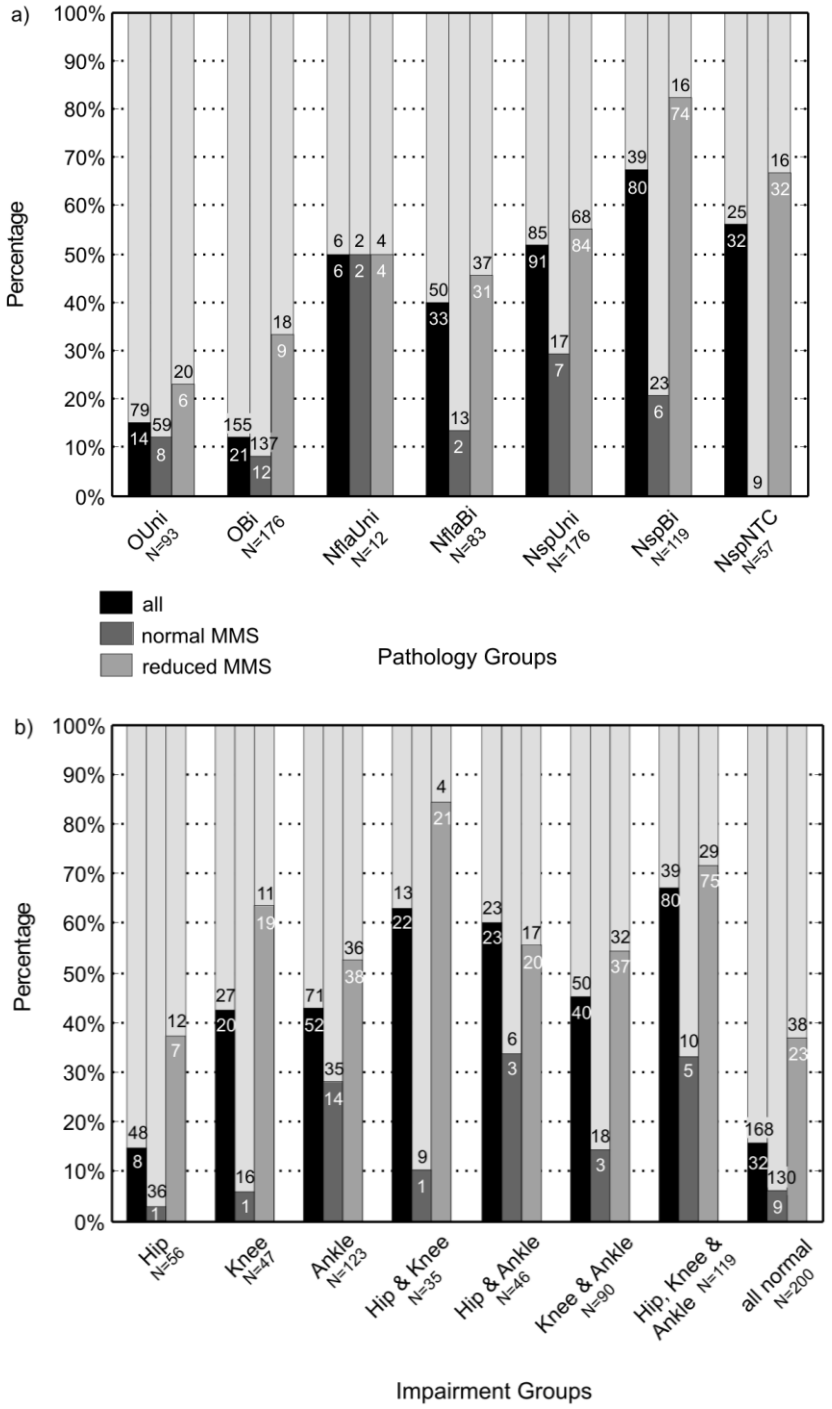

Figure 4.1: Prevalence of premature m. gastrocnemius medialis activity.
The figure shows the number of patients in each pathology group (1a) / impairment group (1b) with abnormal (bottom part) and normal EMG (upper part) expressed in percentage (y-axis) of the total number (in bars) of patients in this group. The first column in each figure represents the distribution across all patients within this group (all). The second column displays the distribution in the subgroup with mean manual muscle strength (MMS) ≥4.5 (almost no rmal MMS), and the third (reduced MMS) shows the distribution in the subgroup with MMS <4.5. The patient groups in Figure 4.1a are decoded as follows: O/N = orthopaedic/neurologic, Uni/Bi = unilateral/bilateral involvement, fla/sp = flaccid/spastic muscles, NTC = no thoracal control. In Figure 4.1b the patients are grouped according to the impaired joint, e.g. the group 'hip' includes patients with an abnormal Gait Variable Score (GVS) of the hip.

The correlations between equinus gait and PPF were low to moderate, and they were merely significant for all patients (total), OUni, OBi, and NspBi (Table 4.3).

Table 4.3: Correlation between premature GM activity and equinus gait.
The *phi* correlation coefficients of premature GM activity and equinus gait are presented for the different pathology groups in the second column. The *p*-values for the correlations are listed in column three. Values in bold are considered significant at $p<0.05$. The patient groups are decoded as follows: O/N = orthopaedic/neurologic, Uni/Bi = unilateral/bilateral involvement, fla/sp = flaccid/spastic muscles, and NTC = no thoracal control.

Group	Phi	p
Total	**0.246**	**.000**
OUni	**0.388**	**.000**
OBi	**0.262**	**.001**
NflaUni	-0.333	.248
NflaBi	0.085	.440
NspUni	0.113	.133
NspBi	**0.227**	**.013**
NspBiNTC	-0.008	.952

Across all patients with normal EMG, 79.0% (347/439) did not show an equinus gait, while 21.0% (92/439) did. Of all patients with PPF, a normal foot contact was present in 56.0% (155/277) of patients and an equinus foot contact in 44.0% (122/277). Patients with an equinus gait but without a PPF during loading response showed a significantly ($p=0.001$) higher mean GM activity during terminal swing than patients with a normal foot contact (Figure 4.2). Additionally, they had significantly lower peak TA activity during loading response ($p=0.026$) than in patients with a normal foot contact (Figure 4.2). Both mean GM activity during loading response and mean TA activity during the terminal swing phase did not differ significantly within these two groups (GM: $p=0.209$, TA: $p=0.318$). Patients with a normal foot contact, despite PPF, revealed a higher mean foot absorption power ($p=0.007$) during loading response compared to patients without PPF (Figure 4.3).

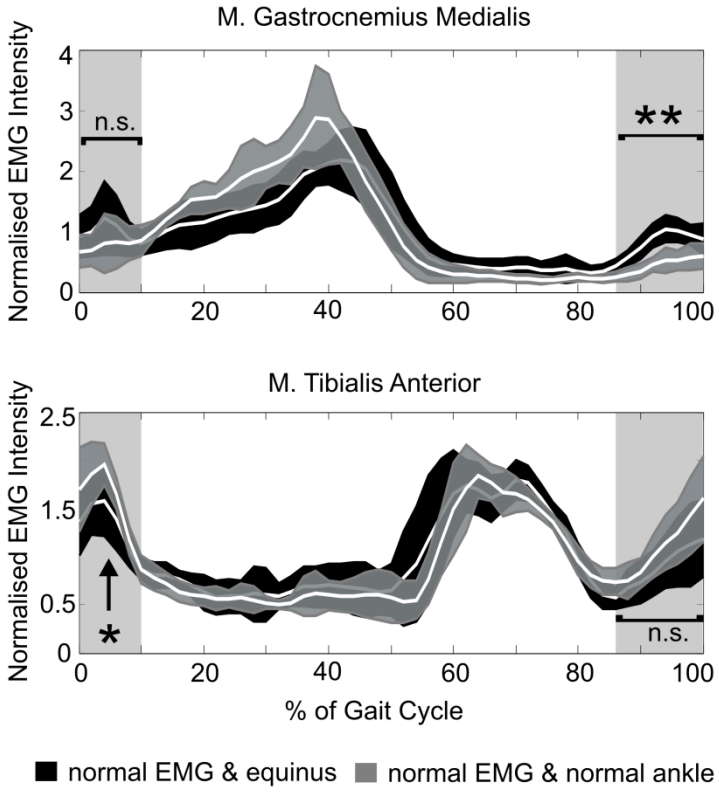

Figure 4.2: Normal foot contact vs. equinus gait in patients with normal EMG.
The EMG of patients with a normal foot contact (grey) and of patients with equinus gait (black) is plotted for an entire gait cycle. The loading response and terminal swing are the gait phases of interest, and are the areas shaded in grey. Presented are the mean and one standard deviation of the enveloped EMG signal for the respective groups. The EMG signals was normalised to the mean amplitude of the signal before they were averaged. Asterisks indicate statistical significant differences (* $p<0.05$, ** $p<0.01$). The abbreviation "n.s." stands for non-significant.

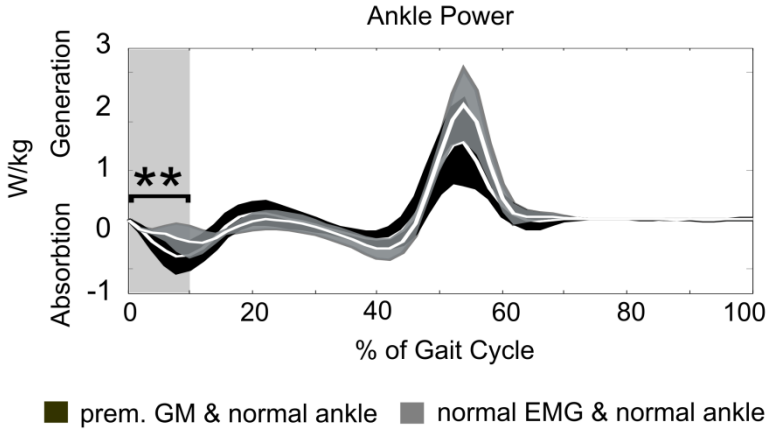

Figure 4.3: Premature gastrocnemius activity vs. normal EMG in patients with normal foot contact.
The ankle power of patients with a normal foot contact and premature m. gastrocnemius medialis (GM) activity (black) and of patients with normal EMG (grey) is plotted for an entire gait cycle. The grey shaded area is the loading response phase. Presented are the mean and one standard deviation of the ankle power for the respective groups. The bold black lines indicate statistical significant differences.

DISCUSSION

This study focused on the association between equinus gait, the muscle strength of patients, and their EMG pattern across a variety of different pathologies. It was hypothesised that muscle weakness is among the causes for PPF, and that PPF correlates with equinus gait. Knowledge on the interrelations between these parameters can assist clinicians in interpreting gait deviations across different patient groups.

In order to prevent overestimation of the number of patients with abnormal muscle timing, the criteria for PPF during loading response were set according to walking speed. Thereby we took into account that the EMG amplitude differs depending on the walking speed [24]. In addition, the activity had to be constantly above the threshold for the entire loading response phase. The detection method for PPF was considered as valid, since none of the healthy controls had an abnormal EMG according to this method.

PPF was present across all pathological groups; hence, PPF is not dependent on one sole neurological component. It remains unclear whether in spastic patients the neurological disease itself is another factor for PPF or whether the higher incidence of PPF is only due to a poorer neuromuscular control. Except for the NflaUni group, the number of patients with PPF increased drastically between the subgroups "almost normal MMS" to "reduced MMS". Although it is difficult to quantify, there exists at least a qualitative relation between the EMG signal and the force of the muscle [25]. Consequently, an explanation for these findings could be that weak patients might need higher muscle activity to produce the same force, or at least a sufficient force to control the joints under a loaded condition. The results for the NflaUni group might have been biased due to a reduced amount of patients. Particularly in the subgroup with normal muscle strength, where there were only four patients.

The formation of patient groups according to their pathology obviously is a limitation. When clustering according to the pathology, inevitably some groups comprised patients with very different diagnoses such as OUni, OBi, NflaBi, whereas other groups, such as NspUni or NspBiNTC, were rather homogenous. The less homogenous groups were composed of patients with very different diagnoses, as the total number of individuals with a given pathology was too small. To account for that, we also grouped the patients according to their impaired joints. Similar to the grouping according to the pathology, weak patients showed PPF more frequently than patients with good muscle strength. Considering that for the two different grouping strategies, the main results were the same, we are confident that the patient group clustering did not bias our work.

The correlations between equinus gait and PPF were lower than expected and often not significant. In the patient groups NflaUni, NflaBi, NspUni, NspBiNTC there seems to exist no such correlation. Even in the patient groups where the correlations were significant (OUni, OBi, NspBi) the *phi* values were low. This fact shows that equinus gait is a predictor for PPF in these patients, however, it is only a weak one. Similarly, PPF and equinus gait are significantly associated for all patients (Total) but only to a low extent. These results can be explained by the unexpected high number of patients with a normal foot contact despite PPF and

also by the unexpected high number of patients with an equinus gait not showing a PPF. In patients without PPF and equinus, the higher GM activity just prior to foot strike, together with lower TA activity could promote equinus gait with lower GM activity needed during the loading response phase of walking. Further, it is possible that these patients use their soleus muscle more to keep the equinus upright. Unfortunately, there were no EMG data on the soleus muscle available. Patients with normal EMG and equinus may still have a higher activity of the GM in loading response, but this activity is not constant or above the thresholds to be categorized as PPF. However, after looking at the mean EMG signals of these patients, this seems to be the case. Patients visually show more GM activity during loading response than patients with a normal foot contact but the difference is not significant. Higher GM activity around foot strike in equinus gait is in accordance with the literature [5,9,11]. Increased activity is supposed to be essential in order to keep the same force generating capacity of the plantarflexor muscles while they are acting on a less-optimal force-length condition [3]. Patients with PPF and a normal foot contact produced higher ankle absorption power than patients with normal EMG and normal foot contact in loading response. An explanation could be that patients with PPF could possibly use their muscle activity to prevent the tibia in translating forward; hence, they might control the knee using PPF during loading response. When this hypothesis can be verified by muscle modelling, this would hold evidence that the plantarflexion-knee extension couple does not only act in mid stance [10] but can be used also during loading response.

CONCLUSION

This study indicates that muscle strength is an aetiological factor for PPF independent of the primary pathology. Even in neurological patients, it is not only spasticity which leads to PPF, but also muscle weakness. In consequence, we conclude that PPF should be regarded as a secondary gait deviation with clinical relevance in all patients. While, for equinus gait, increased GM activity just prior to foot strike or during loading response seems essential, PPF does not necessarily produce an equinus gait. Rather it can also be used to control the knee through the plantarflexion-knee extension couple in loading response.

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CHAPTER 5

THE EFFECT OF TOE WALKING ON THE UPPER BODY

Upper Body Movements in Hemiplegic Children Walking with and without Ankle-Foot Orthosis

Katrin Schweizer, Reinald Brunner & Jacqueline Romkes

Submitted

ABSTRACT

Background: Although both the lower and upper body of patients with hemiplegic cerebral palsy (hemi-CP) are affected, studies on deviated trunk and arm movements during gait are scarce. The aim was to document gait deviations in the upper body of children with hemi-CP walking barefoot (toe walking) and with a hinged ankle-foot orthosis (heel-toe gait).

Methods: Children with hemi-CP walking barefoot and with a hinged ankle-foot orthosis were compared to healthy children. Kinematics of the trunk and upper limbs were investigated. A gap between the 95% confidence intervals defined significant differences.

Findings: Range of motion (RoM) of the pelvis, spine, and thorax tilt was increased in patients compared to controls. In the coronal plane, the pelvis was lower on the affected side within the patients, and they showed a significant increased RoM of the thorax. The pelvis and thorax were more anterior rotated on the unaffected side, and thorax RoM was increased in comparison to the controls. The orthoses had no effect on the trunk movement alterations. Despite both sides are within normal ranges, the unaffected elbow and shoulder flexion RoM was significantly increased compared to hemiplegic side with orthotics.

Interpretation: Trunk kinematics of patients revealed abnormalities in all three planes compared to controls. A hinged ankle-foot orthosis restoring the first ankle rocker had no clear influence on the upper body kinematics. None of the observed gait deviations in the trunk and arms seemed to be a secondary deviation caused by toe walking and lacking of the first ankle rocker. The unaffected arm compensated for the hemiplegic side by increased arm swing.

INTRODUCTION

Patients with cerebral palsy (CP) are the most commonly observed patients in gait laboratories [1,2]. Contrarily to patients with stroke, the brain damage in CP patients occurs prenatal or in early childhood [3,4]. Hemiplegic cerebral palsy (hemi-CP) is one of the subgroups. These patients show involvement of the arm and leg of mainly body side. The neuromuscular impairment is typically of spastic nature.

Due to the unilateral impairment, hemi-CP patients demonstrate an asymmetric leg swing with a 5.5% increased amplitude on the sound side [5]. According to Wren et al. [6], 64% of hemi-CP patients have an equinus gait, 56% a stiff knee, 54% show in-toeing while walking, 48% have excessive hip flexion, and 47% show a crouch gait pattern.

Although, both the upper and lower body side is affected, studies on the trunk and arms in hemi-CP patients are scarce [7]. Hemiplegic stroke patients were reported to have larger lateral displacement of the trunk with accentuation towards the sound side [8]. This was supported by Hsue et al. [9] who found the centre of mass (CoM) displacement to be higher in the mediolateral and vertical amplitude in children with hemi-CP. Riad et al. [7] reported a decreased range of motion (RoM) in the elbow and shoulder on the hemiplegic side, together with an increased flexion of the elbow. Their results are in line with Meyns et al. [5] who found 22.9% reduced arm swing on the involved side compared to healthy children. Further, they found the sound side to compensate by a 53.3% higher arm amplitude. This enhanced arm swing seemed to be driven by trunk rotation towards the unaffected side [5].

As equinus gait is one of the most typical gait deviations in hemiplegia, hinged ankle-foot orthoses (hAFO) are often prescribed in these patients. An hAFO blocks excessive plantarflexion in swing while allowing dorsiflexion. Numerous studies have confirmed the gait improving capabilities of hAFOs for the lower body in hemi-CP patients [10-13]. Patients walked with increased speed [11-13], longer stride and step length [10-13], and improved single support time [12] when wearing an hAFO compared to the barefoot condition. Most importantly hAFOs were found to reduce plantarflexion, especially at initial contract and during mid-swing [10,12,13], thereby restoring a heel-toe gait. At the knee hAFOs decreased the flexion at initial contact [12], and prevented hyperextension in stance [10]. At the hip the range of motion was increased and adduction was reduced compared to barefoot gait [11]. When wearing an hAFO pelvic obliquity was more symmetric [11].

While many studies have concentrated on the effect of hAFOs on the lower body in hemi-CP patients [10-13], similar studies for the upper body are scarce. Patients walking with a posterior leaf spring orthosis revealed increased RoM of the spine and thorax in frontal and transversal plane [14]. Degelaen et al. [15] gave indications on increased trunk motion when hemiplegic patients walked with an ankle-foot orthosis. However, the differences seem not tested statistically. Brunner et al. [11] reported a visual trend of a less pronated arm and wider swing when walking with hAFOs.

Spasticity of the upper body seems to restrict the lower body when walking. Treating this spasticity, by means of botulinum toxin injections, improved gait speed [16] and stride time of the paretic leg in stroke patients [17].

If treating the upper body can improve the gait parameter in the lower body the question arises, whether inversely, treatment of the lower body can improve movement parameters of the upper body as well? However, there was no comprehensive literature on upper body kinematics in hemi-CP patients. Therefore, the parameters that differ from normal when patients walk barefoot with their typical toe walking pattern needed to be identified first. In a second step, it was evaluated how these deviations changed as a result to a heel-toe gait pattern when wearing a hAFO. Trunk movements are considered as an essential component of effective gait [8]. To know which upper body angles are improved by a hAFO is of clinical relevance as it helps discriminating primary from secondary deviations.

METHODS

Participants

For this retrospective study all hemi-CP patients in our gait database from 2006 till 2013 were considered. The patients had to meet the following inclusion criteria: hemiplegia of type CP, aged between 8-18 years, no botulinum toxin-A treatment within the last three months, full-body gait analysis data of barefoot walking and with a hAFO with shoes at the same visit in the gait laboratory, no other assistive devices such as crutches or posterior walker. Further, patients had to show a flat-foot or toe initial contact on the affected side when walking barefoot that was corrected to a heel strike by the hAFO. These enclosing criteria were met by 23 patients. The severity of the affection of the hemiplegic side was rated according to the four gait pattern groups described by Winters et al. [18].

A group of 17 healthy children provided the reference gait data. All participants signed an informed consent. The study was approved by the local ethical committee.

Kinematic Data Collection

Patients and controls walked barefoot at a self-selected speed on a 10 m level ground walkway. Reflective markers (14mm diameter) were attached bilaterally to bony landmarks on the skin of the subjects (Figure 5.1A). Movement of the subjects were tracked by a VICON motion analysis system (six-camera system 460, 120 Hz, years 2006-2010; twelve-camera system MXT20, 200 Hz, since 2011). The Helen Hayes Marker set [19] was used to model the lower body. For the upper body the Plug-in-Gait model (VICON) was applied as described by Gutierrez et al. [20]. Subjects' height, weight, leg lengths, anterior superior iliac spine to trochanter distances, tibial torsions, width of ankles, knees, elbows, wrists, and hands, and the shoulder offsets were measured for individual anthropometric scaling of the model. The knee

alignment device was applied in the static trial to estimate the knee flexion axis (Motion Lab Systems Inc., Los Angeles, USA).

The patient completed a second walking session with a hAFO on the affected side and shoes. Both conditions, barefoot and with a hAFO, were tested on the same day. All but the toe and heel markers remained at the same positions in both conditions. Toe and heel markers had to be attached to the shoes in the hAFO session, and a new static trial was recorded. The hAFOs were hinged, allowing ankle dorsiflexion while blocking plantarflexion (Figure 5.1B). The foot plate of the hAFOs included the entire length of the foot to the tip of the toes, and the posterior part of the hAFOs extended to just below the knee. All hAFOs were custom made to suit the individual needs of each patient and to provide the best possible fit. The hAFO was fitted to the equinus deformity of the foot so that the sole of the shoes and the tibia stood orthogonal. The patients wore the hAFO in daily life; hence, they were accustomed to it.

In addition, RoM of the knee and ankle as well as manual muscle strength [21] were clinically examined. The muscle strength was tested of the knee and ankle flexors and extensors on a scale from 0 (paralysed muscle) to 5 (strong/normal).

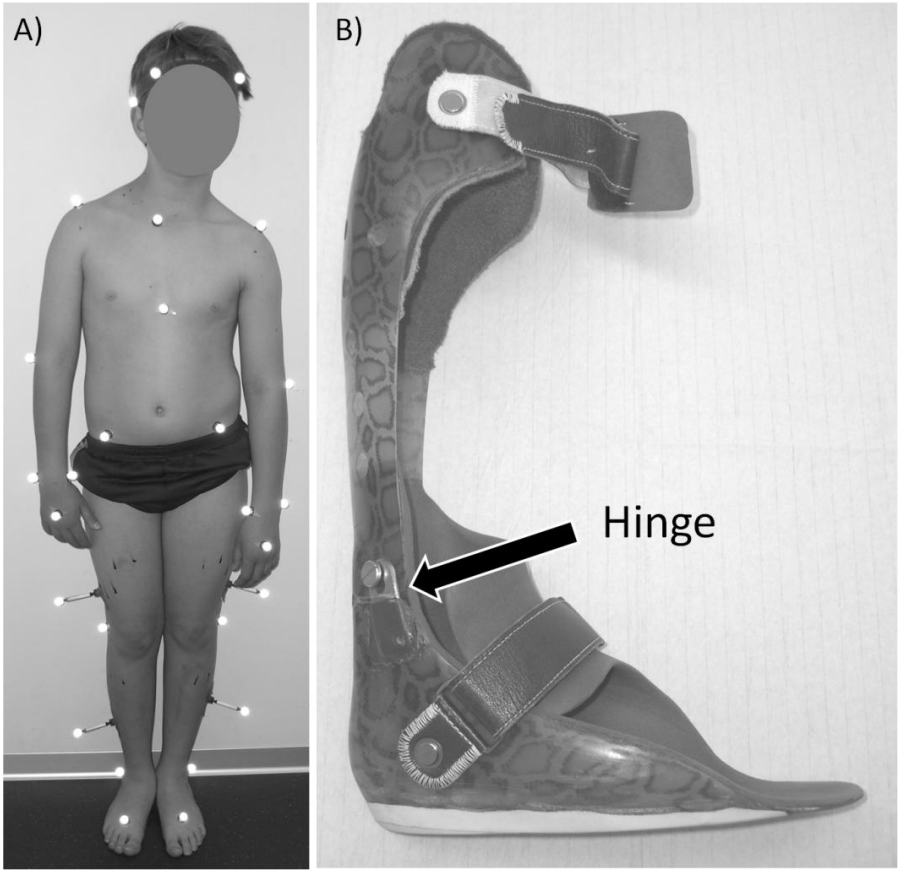

Figure 5.1: Marker placement and orthosis.
Figure 5.1A) pictures the marker placement on a hemiplegic patient. Figure 5.1B) shows a typical hinged ankle-foot orthosis.

Data Analysis

The VICON-software was used for the pre-processing of the data. This included the visual setting of gait events, and filtering of the kinematic data with the built-in Woltring filter (mean squared error set to 10).

The data were post-processed with the MATLAB software (MathWorks Inc., Version R2010a, Natick, MA, USA). All joint angles were time normalised to stance (0-60%) and swing phase (61-100%), and formed together a gait cycle consisting of 101 data points. A gait cycle was defined as the time between two consecutive foot strikes of the same leg. Spatio-temporal parameters were transformed into non-dimensional parameters, accounting for individual anthropometry according to Hof et al. [22].

As upper body kinematics are more variable within subjects than lower body kinematics averaging the trials per subject would distort the data. Therefore, the selection method for a representative trial (SMaRT) [23] was applied to automatically extract one trial for each patient and each control. The method computes the distance of each principal component score to the median of all trials for each angle, and selects the trial that is closest to the median of all trials across all angles. However, the input angles for SMaRT differed from those previously described in Schweizer et al. [23]. In this study the elbow flexion, shoulder flexion and abduction, and the thorax and pelvic angles in all three planes and both body sides were used as input. The representative trial was chosen from 7 trials on average (patient range: 3-14, control range: 3-15). For further analysis one body side in healthy subjects was selected randomly, in patients the affected and unaffected side were compared.

The variables of interest were the following parameters of the joint angles in sagittal plane: ankle at initial contact, mean in stance, maximum and mean in swing; RoM and mean over the gait cycle of the pelvis, spine, thorax, shoulder and elbow. In coronal plane: maximum in stance and minimum in swing for the pelvis and spine; minimum in stance and maximum in swing for the thorax; additionally, RoM and mean over the gait cycle for the pelvis, spine, thorax, and shoulder. In transversal plane the variables of interest were: the RoM and mean over the gait cycle of the pelvis, spine, and thorax.

Statistic

The 95% confidence intervals (CI) for the RoM, muscle strength, spatio-temporal parameters and joint angle parameters were calculated. A parameter of which the CI within the patients or compared to the controls did not overlap was defined as significantly different with a 95% probability [24,25].

RESULTS

The control group and patient group had similar age and anthropometrics Table 5.1. Especially the RoM in dorsiflexion of the ankle and the muscle strength of the plantarflexors and dorsiflexors on the hemiplegic side were severely reduced compared to the unaffected side (Table 5.1).

Walking speed did not differ neither between patients and controls nor between the two walking condition. When walking barefoot, the patients showed earlier foot-off on the affected side compared to the controls. Foot-off was earlier and step time was longer on the affected side than on the unaffected side. When walking with a hAFO, foot-off on the unaffected side came later than in controls. Analogue to the barefoot condition, step time was longer and foot-off earlier on the unaffected compared to the affected side (Table 5.1). The step time was also longer compared to the controls.

The mean joint angles for the patients are plotted in Figure 5.2. Pelvis and thorax angles are given in absolute angles referring to the gait laboratories' axis, and the spine angles are relative angles between those two segments.

Barefoot Walking

When walking barefoot, the affected ankle in *sagittal plane* was more plantarflexed at initial contact, during swing and stance than on the unaffected side and in the control group (Table 5.2). The maximum plantarflexion in swing of the hemiplegic side was higher than on the unaffected side.

In the upper body, the RoM of the pelvis, spine, and thorax was increased in patients compared to the controls. The difference for an excessive anterior pelvic tilt and a posterior spine tilt of the patients narrowly misses significance. Similarly, the RoM of the shoulder, in spite of seeming to be clinically increased on the unaffected compared to the affected side, was not significantly different. The elbow was significantly more flexed on the hemiplegic side in comparison to the unaffected side and the controls. The same tendency was seen at the unaffected elbow, although without significance.

The pelvis was lower on the affected side than on the non-affected side in the *coronal plane*. Compared to the controls, the patients showed a significant increased RoM of the thorax on the affected side. The increase of this parameter on the unaffected side was not significant. A tendency of higher shoulder RoM of both arms in patients was observed.

In the *transversal plane*, the pelvis and thorax were more internally rotated on the unaffected side. The RoM of the thorax was increased on both sides of the patients in comparison to the controls. On the affected side, the pelvis, spine, and thorax were externally rotated compared to the unaffected side, the pelvic also more than the controls.

Walking with hAFO

Wearing hAFOs reduced the excessive plantarflexion at initial contact and during swing phase (Table 5.2). Nevertheless, plantarflexion at initial contact and during stance was still increased compared to the unaffected side and to the controls. Maximum plantarflexion in swing was reduced compared to the unaffected side and controls.

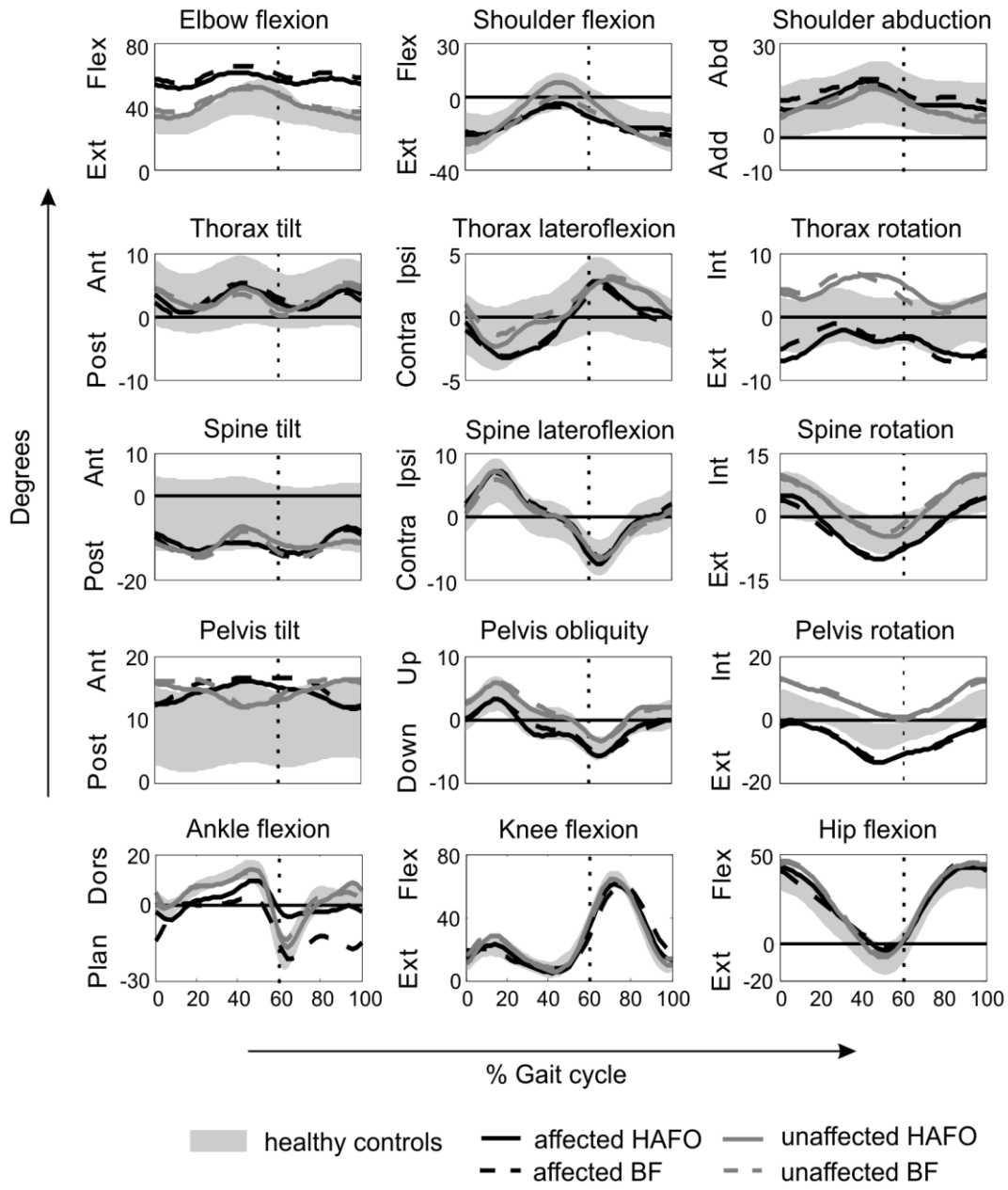

Figure 5.2: Joint angles.
Mean joint angles in degrees of the patients are plotted normalised to a gait cycle (y-axis). The hemiplegic side of the patients is black, the unaffected side grey. Dashed lines illustrate barefoot walking (BF), continuous lines represent walking with hinged ankle-foot othrosis (hAFO). The grey band displays the mean ± one standard deviation of the control group. All data were normalised to stance and swing phase separately, with toe-off at 60% of the gait cycle which is indicated by the dashed vertical line.

Table 5.1: Subjects' characteristics.
Reported are means ± one standard deviation (95% confidence interval) unless stated otherwise. Significant differences are highlighted in bold and the direction of the difference is indicated by arrows.

Parameter	Controls (n=17)	Patients (n=23)	
Age in years (range)	12.8 (8-18)	12.4 (8-18)	
Height [m]	1.59 ±0.14 (1.51, 1.66)	1.49 ±0.12 (1.43, 1.54)	
Weight [kg]	47.8 ±10.7 (42.1, 53.4)	42.1 ±13.6 (36.1, 48.1)	
Sex [female/male]	8/9	9/14	
Hemiplegic type [type 1/2/3]	-	15/5/3	
		Unaffected side	Affected side
Analysed side [left/right]	9/8	11/12	12/11
Knee ext.,hip 90° flex. [°]*	-	-39 ±11 (-44, -34)	-46 ±10 (-50, -42)
Knee ext., hip ext. [°]*	-	6.5 ±3.1 (5.1, 7.9)	**2.6 ±4.4 (0.7, 4.6) ↓**
Dorsiflex, knee 90° flex. [°]*	-	17.2 ±4.1 (15.4, 19.0)	**-0.4 ±10.2 (-4.9, 4.1) ↓**
Dorsiflex, lower ankle joint fixed [°]*	-	8.0 ±4.4 (4.3, 11.8)	**-8.0 ±12.4 (-13.5, -2.6) ↓**
MMS knee flex.	-	5.0 ±0.1 (4.9, 5.0)	**4.5 ±0.6 (4.2, 4.8) ↓**
MMS knee ext.	-	5.0 ±0.1 (5.0, 5.0)	**4.7 ±0.3(4.5, 4.8) ↓**
MMS active knee ext. deficit [°]	-	0.9 ±3.2 (-0.5, 2.3)	2.4 ±4.7 (0.3, 4.4)
MMS plantarflexion stand-ing	-	4.9 ±0.3 (4.8, 5.0)	**3.3 ±1.4 (2.7, 4.0) ↓**
MMS dorsiflex.	-	4.9 ±0.2 (4.9, 5.0)	**3.1 ±0.9 (2.7, 3.5) ↓**

		Barefoot unaffected	hAFO unaffected	Barefoot affected	hAFO affected
Walking speed [ND]	0.45 ±0.05 (0.43, 0.48)	0.42 ±0.05 (0.40, 0.44)	0.45 ±0.06(0.43, 0.48)	0.43 ±0.05 (0.40, 0.45)	0.45 ±0.06 (0.43, 0.48)
Cadence [ND]	34 ±2 (33, 35)	34 ±3 (33, 35)	33 ±2 (32, 34)	34 ±3 (32, 35)	32 ±2 (32, 33)
Step time [ND]	1.76 ±0.14 (1.69, 1.84)	1.67 ±0.16 (1.60, 1.74)	1.78 ±0.15(1.71, 1.84)	**1.90 ±0.18 (1.82, 1.98) ↑**	**1.93 ±0.13 (1.87, 1.99)↖↑**
Step length [ND]	0.79 ±0.07 (0.75, 0.83)	0.75 ±0.09 (0.71, 0.79)	0.83 ±0.10 (0.79, 0.87)	0.76 ±0.08 (0.72, 0.79)	0.83 ±0.10 (0.79, 0.87)
Foot-off [% of gait cycle]	60.1 ±1.9 (59.1, 61.2)	61.8 ±2.4 (60.8, 62.9)	**62.3 ±2.2 (61.3, 63.3)↖**	**57.2 ±2.0 (56.3, 58.1) ↙↓**	**58.7 ±2.0 (57.9, 59.6) ↓**

Hemi type = Classification according to Winter et al. (1987)

* = Joint mobility measures. Negative values indicate a deficit to reach neutral zero position.

MMS = Manual muscle strength in clinical testing

hAFO = Hinged ankle-foot orthosis condition

ND = Non-dimensional values according to Hof et al. (1996)

↖↙ = Higher/reduced compared to control group

↑↓ = Affected side higher/reduced than unaffected side

Table 5.2: Joint angle parameters.
Reported are means ± one standard deviation (95% confidence interval). Significant differences are highlighted in bold and the direction of the differences is indicated by arrows.

Body plane	Joint angle	Gait phase	Para-meter	Controls	Patients unaffected side		Patients affected side	
					Barefoot condition	hAFO condition	Barefoot condition	hAFO condition
Flexion/ Tilt Positive: Planar-flexion/ Anterior/ Flexion	Ankle	IC		-1.9 ±4.3 (-4.1, 0.3)	-0.9 ±6.1 (-3.6, 1.7)	-5.2 ±5.0 (-7.3, -3.0)	**14.2 ±7.1 (11.1, 17.3) ↖↑**	**2.8 ±4.8 (0.8, 4.9) ↖↑↴**
		St	Mean	-6.0 ±2.5 (-7.3, -4.8)	-6.0 ±4.0 (-7.7, -4.3)	-6.4 ±4.3 (-8.3, -4.6)	**1.7 ±7.6 (- 1.6, 4.9) ↖↑**	**-2.2 ±5.2 (-4.4, 0.1) ↖↑**
		Sw	Max	19.5 ±6.9 (16.0, 23.0)	14.5 ±9.0 (10.6, 18.4)	17.1 ±6.5 (14.3, 19.9)	**24.3 ±10.8 (19.6, 29.0) ↑**	**5.1 ±5.4 (2.7, 7.4) ↙↓↴**
			Mean	3.0 ±4.3 (0.8, 5.2)	1.2 ±5.5 (-1.2, 3.6)	1.8 ±4.9 (-0.3, 3.9)	**16.2 ±8.9 (12.3, 20.0) ↖↑**	**2.6 ±4.8 (0.5, 4.7) ↴**
	Pelvis	GC	RoM	3.6 ±1.2 (3.0, 4.2)	**7.0 ±3.1 (5.7, 8.4) ↖**	**7.0 ±3.1 (5.6, 8.3) ↖**	**7.5 ±3.3 (6.1, 9.0) ↖**	**6.6 ±3.4 (5.1, 8.1) ↖**
			Mean	9.1 ±6.2 (5.9, 12.2)	15.0 ±6.7 (12.1, 17.9)	14.3 ±6.7 (11.4, 17.1)	15.1 ±6.6 (12.2, 18.0)	14.1 ±6.8 (11.2, 17.0)
	Spine	GC	RoM	5.0 ±1.7 (4.1, 5.9)	**10.3 ±5.3 (8.0, 12.6) ↖**	**9.2 ±5.3 (7.0, 11.5) ↖**	**10.2 ±5.6 (7.8, 12.6) ↖**	**8.6 ±5.6 (6.2, 11.0) ↖**
			Mean	-5.0 ±8.1 (-9.2, -0.9)	-12.4 ±10.0 (-16.8, -8.1)	-11.4 ±8.9 (-15.3, -7.6)	-12.3 ±9.8 (-16.6, -8.1)	-11.5 ±8.6 (-15.2, -7.8)
	Thorax	GC	RoM	4.0 ±1.0 (3.5, 4.5)	**7.4 ±2.9 (6.2, 8.6) ↖**	**6.8 ±2.7 (5.6, 8.0) ↖**	**7.3 ±3.3 (5.8, 8.7) ↖**	**6.8 ±2.8 (5.6, 8.0) ↖**
			Mean	3.1 ±4.8 (0.6, 5.5)	2.7 ±4.6 (0.7, 4.7)	2.8 ±5.0 (0.7, 5.0)	2.9 ±4.4 (1.0, 4.8)	2.6 ±4.9 (0.5, 4.7)
	Shoul-der	GC	RoM	25.8 ±17.8 (16.6, 34.9)	29.0 ±16.1 (22.1, 36.0)	37.7 ±15.8 (30.9, 44.5)	20.9 ±11.3 (16.0, 25.8)	**19.2 ±9.9 (14.9, 23.5) ↓**
			Mean	-9.0 ±5.7 (-12.0, -6.1)	-13.5 ±7.9 (-16.9, -10.1)	-10.5 ±7.9 (-13.9, -7.1)	-14.6 ±6.1 (-17.3, -12.0)	-13.6 ±6.2 (-16.3, -10.9)
	Elbow	GC	RoM	19.4 ±14.3 (12.0, 26.7)	23.9 ±11.1 (19.1, 28.7)	25.4 ±10.8 (20.7, 30.1)	16.8 ±8.8 (13.0, 20.6)	**15.5 ±8.8 (11.7, 19.3) ↓**
			Mean	35.6 ±5.5 (32.7, 38.4)	42.8 ±10.6 (38.2, 47.4)	41.1 ±10.5 (36.6, 45.6)	**59.9 ±19.0 (51.6, 68.1) ↖↑**	**56.1 ±15.8 (49.3, 63.0)↖↑**
Obliquity/ Abduction Positive: Up/ abduc-tion	Pelvis	St	Max	4.5 ±2.3 (3.3, 5.7)	6.2 ±3.1 (4.9, 7.6)	6.1 ±3.0 (4.8, 7.4)	3.6 ±3.2 (2.2, 5.0)	**3.4 ±3.0 (2.1, 4.7) ↓**
		Sw	Min	-4.8 ±1.8 (-5.7, -3.8)	-3.7 ±4.0 (-5.4, -1.9)	-3.5 ±3.2 (-4.9, -2.1)	-5.8 ±2.7 (-7.0, -4.6)	**-6.2 ±2.7 (-7.3, -5.0) ↓**
		GC	RoM	9.3 ±3.1 (7.7, 10.9)	10.1 ±4.0 (8.4, 11.8)	9.9 ±4.1 (8.1, 11.6)	9.5 ±3.6 (7.9, 11.0)	9.6 ±4.0 (7.9, 11.4)
			Mean	-0.3 ±1.6 (-1.1, 0.6)	1.4 ±2.6 (0.3, 2.5)	1.3 ±2.0 (0.4, 2.2)	**-1.2 ±2.2 (-2.2, -0.3) ↓**	**-1.4 ±1.9 (-2.2, -0.6) ↓**

Obliquity/ Abduction Positive: Up/ abduction	Spine	St	Max	6.0 ±3.4 (4.2, 7.7)	6.3 ±3.9 (4.6, 8.0)	7.7 ±4.0 (5.9, 9.4)	7.3 ±5.6 (4.9, 9.7)	7.5 ±5.5 (5.1, 9.9)
		Sw	Min	-6.7 ±2.8 (-8.2, -5.3)	-7.1 ±5.7 (-9.5, -4.6)	-7.2 ±5.6 (-9.6, -4.8)	-6.8 ±4.9 (-8.9, -4.7)	-7.9 ±4.2 (-9.7, -6.0)
		GC	RoM	12.7 ±3.6 (10.9, 14.6)	13.8 ±4.4 (11.8, 15.7)	14.9 ±4.0 (13.2, 16.7)	14.1 ±4.8 (12.1, 16.2)	15.4 ±4.7 (13.3, 17.4)
	Thorax	St	Min	-2.0 ±2.5 (-3.3, -0.7)	-2.1 ±3.4 (-3.6, -0.6)	-3.0 ±4.1 (-4.8, -1.2)	-4.0 ±3.4 (-5.5, -2.5)	-4.1 ±3.4 (-5.6, -2.7)
		Sw	Max	2.0 ±2.9 (0.5, 3.5)	3.8 ±4.0 (2.1, 5.6)	3.8 ±3.9 (2.1, 5.5)	2.7 ±4.7 (0.7, 4.8)	3.4 ±3.9 (1.7, 5.1)
		GC	RoM	4.4 ±2.6 (3.0, 5.7)	6.9 ±3.9 (5.3, 8.6)	7.2 ±4.3 (5.3, 9.1)	**7.3 ±3.8 (5.7, 9.0) ↖**	**8.1 ±3.7 (6.4, 9.7) ↖**
			Mean	-0.1 ±2.1 (-1.1, 1.0)	0.9 ±2.9 (-0.4, 2.1)	0.4 ±3.2 (-0.9, 1.8)	-0.7 ±3.3 (-2.1, 0.7)	-0.4 ±2.8 (-1.7, 0.8)
	Shoulder	GC	RoM	8.2 ±6.4 (4.9, 11.5)	13.0 ±6.8 (10.1, 16.0)	13.5 ±5.9 (10.9, 16.0)	12.1 ±6.7 (9.1, 15.0)	12.3 ±8.8 (8.5, 16.1)
			Mean	10.9 ±8.5 (6.5, 15.2)	10.9 ±6.4 (8.1, 13.6)	9.9 ±6.0 (7.2, 12.5)	14.3 ±9.4 (10.2, 18.4)	12.1 ±8.5 (8.4, 15.8)
Rotation Positive: Internal	Pelvis	GC	RoM	12.5 ±7.0 (8.9, 16.1)	16.1 ±6.8 (13.1, 19.0)	16.0 ±6.7 (13.1, 18.9)	16.3 ±6.7 (13.3, 19.2)	15.8 ±6.2 (13.1, 18.5)
			Mean	-0.4 ±2.2 (-1.5, 0.8)	**6.2 ±4.4 (4.3, 8.1) ↖**	**6.4 ±4.1 (4.6, 8.1) ↖**	**-6.6 ±3.7 (-8.2, -5.0) ↙↓**	**-7.0 ±4.0 (-8.7, -5.2) ↙↓**
	Spine	GC	RoM	11.1 ±5.4 (8.3, 13.9)	15.2 ±5.0 (13.0, 17.3)	**16.5 ±5.4 (14.2, 18.8) ↖**	16.1 ±6.4 (13.3, 18.8)	**16.5 ±5.6 (14.1, 18.9) ↖**
			Mean	0.5 ±3.7 (-1.4, 2.4)	3.1 ±3.7 (1.5, 4.7)	2.8 ±3.4 (1.3, 4.2)	**-3.0 ±4.0 (-4.7, -1.3) ↓**	**-2.8 ±3.4 (-4.3, -1.3) ↓**
	Thorax	GC	RoM	6.1 ±2.4 (4.8, 7.3)	**9.9 ±4.1 (8.1, 11.6) ↖**	**11.0 ±4.0 (9.3, 12.8) ↖**	**9.5 ±3.2 (8.2, 10.9) ↖**	**10.1 ±4.5 (8.2, 12.1) ↖**
			Mean	-0.9 ±3.5 (-2.7, 0.9)	**3.7 ±4.4 (1.8, 5.6) ↖**	**4.0 ±4.3 (2.1, 5.8) ↖**	**-4.1 ±4.2 (-5.9, -2.3) ↓**	**-4.5 ±4.4 (-6.4, -2.6) ↓**

IC = Initial contact

St = Stance phase

Sw = Swing phase

RoM = Range of Motion

hAFO = Hinged ankle-foot orthosis condition

GC = Gait cycle

↖↙ = Higher/reduced compared to control group

↑↓ = Affected side higher/reduced than unaffected side

↴↥ = Reduced/higher than in barefoot condition

All parameters that were abnormal in the upper body when walking barefoot were unchanged with hAFOs. On the affected side, the pelvis was less upward tilted in stance and more downward in swing compared to unaffected side, it was still within the norm though. RoM of spine rotation with hAFO was higher than in the controls. The difference between the unaffected and affected elbow and shoulder swing increased significantly, but both sides were within the range of the controls. None of the upper body kinematic parameters of the same side were different between the barefoot and hAFO condition.

DISCUSSION

The primary objective of this study was to evaluate trunk and arm movements in children with hemi-CP during gait. It was investigated which kinematic upper body parameter in patients differed from the norm when walking barefoot. Additionally, the influence of a hAFO on the upper body during gait was examined. It is of high clinical relevance to understand which upper body parameters are improved by wearing a hAFO, as these parameters are secondary deviations due to toe walking and not primary due to spasticity.

Patients showed a typical toe walking pattern on the hemiplegic side with increased plantarflexion at initial contact and during swing phase of gait when walking barefoot. In this specific group of hemi-CP patients the hAFO restored the initial heel contact and adequate foot clearance during swing. However, as the hAFO is designed to block plantarflexion, patients are limited in their push-off around toe-off where plantarflexion would be needed. This was verified by the reduced ankle plantarflexion in swing which indicates a reduced power generation at push-off [14].

The increased RoM in the trunk and shoulders in sagittal and coronal plane indicate a more unstable gait [14]. Thorax RoM of the controls and hemiplegic patients in the barefoot condition were similar to the reported values by Molenaers et al. [14]. Patients walked with increased lordosis due to excessive pelvic anterior tilt. The transverse plane kinematics revealed that the trunk is rotated so that the unaffected side is in front; hence, it is anterior rotated in both conditions.

The hAFO did not have a strong effect on the trunk motion. Although the first ankle rocker was restored, no clinically relevant changes compared to the barefoot condition were observed in the trunk. As the increased RoM in the trunk did not improve by the hAFO, it does not seem to be caused by the toe walking gait pattern in the barefoot condition. A possible explanation could be that lateral trunk movements usually compensates for hip abductor weakness. However, hAFOs cannot control for hip abductor weakness, as they only control the foot mainly in sagittal plane.

Contrarily, Molenaers et al. [14] found significantly increased thorax RoM when wearing an orthosis. However, these results are not directly comparable as diplegic and hemiplegic patients were mixed and wore posterior leaf spring orthoses. Another explanation for the divergence of the results is that in the study of Molenaers et al. [14] patients walked significantly

faster and with increased step length when wearing an orthosis. In our study, these parameters did not differ significantly although they increased slightly in the hAFO condition.

The observed asymmetry between the affected and unaffected arm swing in the elbow and shoulder is in line with Meyns et al. [5] and with the more severe hemiplegic group of Riad et al. [7]. The asymmetry increased further when wearing a hAFO. A possible explanation can be that the increased arm swing on the unaffected side is a compensation to achieve slightly faster walking speeds with hAFOs. This explanation is supported by Stephenson et al. [26] who found increased arm swing amplitudes of the non-paretic side in stroke patients when walking faster. Although visual improvement of arm position was reported by Brunner et al. [11], no improvement by hAFO in hemiplegic arm position were observed in the parameters investigated within our study. However, pronation of the arm was not analysed as it was not implemented in the model.

One has to bear in mind that in this study one third of the patients had the mildest type of gait deviation (type 1) according to Winters et al. [18]; hence, it is possible that more severely impaired patients show increased gait deviations in the upper body. Furthermore, all patients were accustomed to walk with hAFOs in daily life. Therefore, it is theoretically possible that they have learned and adapted a gait pattern by wearing hAFOs, which they now use in barefoot walking as well. This is an open question for future research. A limitation of this study is the comparison between the barefoot condition and hAFOs in shoes. Data for a shoes-only condition are missing. However, it is also critical to analyse the patients with shoes only as the same shoes worn with hAFOs are typically too big if worn without hAFO. This could distort the gait pattern. Additionally, each patient wore individualised hAFO. This assured the best possible fit on one hand, on the other hand it implies that the hAFOs were not perfectly similar.

CONCLUSION

We conclude that trunk kinematics of a specific group of hemi-CP patients deviate from normal children in all three planes. Kinematic upper body deviations were observed in both conditions: barefoot walking with a toe contact and when demonstrating a heel-toe gait in a hAFO. The entire trunk was rotated in the patients, so that the unaffected side was anterior rotated over the entire gait cycle. The hemiplegic elbow was almost double as much flexed as in typical developed children. Wearing hAFOs had little to no influence on the upper body kinematics in hemi-CP patients during gait. None of the observed gait deviations in the trunk and arms seemed to be a secondary deviation caused by toe walking or a missing of the first ankle rocker. As described before [5,26] this study supports the impression that the unaffected arm compensates for the hemiplegic side by increased arm swing.

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CONFLICT OF INTEREST STATEMENT

All authors disclose any financial and personal relationships with other people or organisations that could inappropriately influence or bias their work.

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CHAPTER 6

CONCLUSION & OUTLOOK

CONCLUSION

The primary focus of this thesis relied on the principals of pathological gait across different patients. The objective was to confirm that gait deviations are not only a result of the primary disease, but can also originate from muscle weakness. Additionally, it was aimed to differentiate primary deviations from secondary. In the following, the knowledge gained from *Chapters 2-5* is summarised in a conclusion. Furthermore, indications for future research are emphasised.

Selection of a Representative Trial

Chapter 2 established a Method for Selecting a Representative Trial (SMaRT) from multiple measures in gait analysis. This method was applied for all other studies within this thesis. Besides its benefits of selecting a representative trial quickly, it is objective, repeatable, and automatic. SMaRT is robust against a limited number of outliers within the data. Nevertheless, it is advisable to select the input parameters thoughtfully. On one hand, SMaRT should run over all the important parameters which are going to be analysed afterwards. This is because the selected trial might be representative for the input parameters only. On the other hand, the more of the input parameters are used, the more likely it becomes that the selected trial is not representative for each single one of these parameters. Furthermore, it can be unwise to apply SMaRT to datasets that have high within-subject variability. However, this is a general problem of the dataset itself and not a real weakness of SMaRT. In datasets with high within-subject variability, there simply does not exist a characteristic trial. Nonetheless, the algorithm does not evaluate the variability within the data. When applying SMaRT in clinical decision-making or in studies with very few subjects, it is advisable to check the consistency of the data beforehand. Despite the explained constraints, SMaRT is a valuable tool if one wishes to select one trial from a dataset. Other than computing an average over the trials, SMaRT keeps an actually measured trial. Especially, for data driven computer simulations this can be highly desirable. The same applies to studies where the data would be distorted by averaging. Practically, SMaRT is applicable to all types of movement parameters. It can be adapted to any number of input parameters. Therefore, it is generally advantageous for any type of movement analysis curves, even beyond the field of clinical gait analysis.

The Influence of Muscle Strength on Gait Deviations

Comparing patients with different primary diseases is a constructive method to understand which gait deviations are primary and which secondary. However, this has barely ever been done before in the literature. In order to compare different patient groups, one has to know which other parameters, e.g. muscle strength, do have an effect on the walking pattern in these patients. Additionally, it was important to investigate whether the effect of muscle strength on gait deviations behaves similar across different patient groups. *Chapter 3 and 4* have emphasised the importance of muscle strength on the gait pattern of patients. The clinical impression

that the weaker the patients are, the more abnormal they walk was confirmed. This was observed through the kinematic as well as electromyographic parameters.

The kinematic gait deviation, measured by the Gait Profile Score (GPS), raised to a similar extent with increasing muscular weakness across all patient groups. The results from *Chapter 3* led to the conclusion that the response to muscle weakness does not defer between patients with different pathologies. Contrarily, the GPS coefficient at normal muscle strength differed significantly between the various groups. A trend of an increased gait deviation with an increased severity of the disease was also noted. Consequently, the basic pathology adds an offset to the GPS that depends on the severity of the primary affection. This is a possible explanation as to why in neurologic patients, the gait pattern, despite successful surgery hardly ever appears normal.

Muscle weakness is amongst the aetiological factors for abnormal electromyographic timing, such as premature plantarflexor activity (PPF). Independent on whether the patients were grouped according to their pathology or to the impaired joints, PPF was more prevalent in weak patients than patients with normal muscle strength (*Chapter 4*). Therefore, PPF can be understood as a secondary deviation due to its dependency on muscle weakness, at least to some extent. Some patients seemed to use PPF in producing an equinus gait. Others patients might need PPF to control the knee through the plantarflexion-knee extension coupling during the loading response phase. Despite normal muscle strength reducing the prevalence of PPF across all different patient groups, PPF was still present amongst these patients. This provides indications for further factors causing PPF. As this was not the main subject of this thesis, one can only speculate about these factors. The primary pathology could have an influence, as could the malalignment of joints.

In conclusion, it is of high importance to take the muscle strength into account when interpreting gait data. Therapy should not only focus on the primary pathology, but also on increasing the muscle strength of a weak patient, if possible.

Spasticity and Gait Deviations

Originally, one would have expected that the gait of patients with neurologic disease and spastic muscles would deviate more than that of neurologic patients without spasticity. However, this thesis revealed an unexpected low number of differences between patients with diplegia and patients with bilateral neurological diseases and flaccid muscles. This gives implication to question the influence of spasticity to gait deviations. Spasticity seems of minor importance when comparing the lower joint angles of patients with diplegia and neurologic patients with flaccid muscles bilaterally. Contrarily in tetraparetic patients, an increase of the kinematic gait deviations in the lower body were observed. On the basis of the presented data, it is not possible to distinguish whether the higher GPS offset is caused by the increased stiffness or by the lack of trunk control. These two factors are probably linked. Nevertheless, affection of the upper body appears to be an important factor that can increase gait kinematic deviations.

Gait Deviations in the Upper Body of Hemiplegic Patients

Patients with hemiplegic cerebral palsy typically walk with an initial toe contact. Toe walking goes along with a reduced gait stability due to a smaller base of support. Wearing a hinged ankle-foot orthosis corrects the foot to a heel-toe gait and assures contact of the entire foot with the floor. Although, it could have been expected that this would stabilise the gait pattern, walking with an orthosis did not reduce the sway of the trunk to a clinically relevant extent. Concluding from the results in *Chapter 5*, none of the trunk deviations observed in this specific patient group seemed to be a secondary deviation that was caused by the toe walking gait pattern. The unaffected arm tends to compensate for the reduced arm swing in the hemiplegic arm.

OUTLOOK

While studies are designed to dissolve specific research questions, the process of answering these questions can also create new potential for further research. In *Chapter 4* it was hypothesised that premature plantarflexor activity during loading response is used to control the knee. This implies that the plantarflexion-knee extension couple would not only operate during mid-stance phase, but also during loading response. Muscle modelling could verify the explanation to the premature muscle activity in a patient with a normal ankle position during initial contact.

Furthermore, it would be interesting to conduct an intervention study where weak patients undergo muscle strength training. According to the results of this thesis, one would expect the patients with a positive muscle strength outcome to have a lower GPS and less premature gastrocnemius muscle activity than before an intervention. However, this would need approval by an intervention study. If the results would come out as expected, this would prove that the results of this thesis are not only valid when comparing different patients, but also within patients.

In general this thesis emphasises the need for further comparisons of gait deviations in patients with different pathologies, especially in order to understand the effect of spasticity on the gait pattern. The studies within this thesis have shown fewer differences between patients with spasticity and other neurological patients than expected, at least for diplegic patients. However, it is possible that spasticity is more visible in other gait parameters or through different analysis techniques, e.g. wavelet analysis. The questions that can be proposed are: In which gait parameters can spasticity be observed? Which gait deviations are caused by spasticity, and which are solely the result of a muscle weakness?

LIST OF ABBREVIATIONS

2D/3D	Two/three-dimensional
BF	Barefoot
BMI	Body Mass Index
CI	Confidence interval
CoM	Centre of mass
CP	Cerebral Palsy
EMG	Electromyography
GC	Gait cycle
GDI	Gait Deviation Index
GGI	Gillette Gait Index
GM	Gastrocnemius medialis muscle
GPS	Gait Profile Score
GRF	Ground reaction force
GVS	Gait Variable Scores
hAFO	Hinged ankle-foot orthosis
IC	Initial contact
ISw	Initial swing
LR	Loading response
MMC	Myelomeningocele
MMST	Manual muscle strength testing
MSt	Mid stance
MSw	Mid swing
ND	Non-dimensional
NflaBi	Patients with neurologic impairment and flaccid muscles bilateral
NflaUni	Patients with neurologic impairment and flaccid muscles unilateral
NspBi	Patients with neurologic impairment and spastic muscles bilateral
NspBiNTC	Patients with neurologic impairment and spastic muscles bilateral without/no thoracal control
NspUni	Patients with neurologic impairment and spastic muscles uninilateral
OBi	Patients with orthopaedic impairment bilateral
OUni	Patients with orthopaedic impairment uninilateral
PC	Principal Component
PCA	Principal Component Analysis
PPF	Premature plantarflexor activity
PSw	Pre-swing
RoM	Range of motion
SD	Standard deviation
SE	Standard error
SMaRT	Selection Method for a Representative Trial
TA	Tibialis anterior muscle
TSt	Terminal stance
TSw	Terminal swing

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Katrin Schweizer

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About the Author

CURRICULUM VITAE

Personal Information

Name	Katrin Irina Schweizer
Date of Birth	19th May 1984
Place of Birth	Freiburg im Breisgau
Nationality	German

Education

May 2010 - Sept. 2013	PhD Candidate at University of Basel in Sports Science Title of Thesis: Principles of Pathological Gait Supervisors: Prof. Dr. med. Reinald Brunner, Prof. Dr. Bert Müller Tutor: Dr. Jacqueline Romkes
Sept. 2007 – May 2009	Master of Science at the Technical University of Munich in Sports Science with focus on leisure sports and elite sports. Title of Thesis: Force Enhancement during and following strength in multi-joint leg extension. Supervisors: Dr. Daniel Hahn, Prof. Dr. Ansgar Schwirtz
Oct. 2004 – Sept. 2007	Bachelor of Arts at the University of Freiburg in Sports Science with focus on public health, rehabilitation and prevention. Title of Thesis: Innervationsstruktur der wichtigsten Muskeln bei verschiedenen Kippbewegungen im Gerätturnen, Supervisors: Dr. Klaus-Jürgen Müller, Prof. Dr. Albert Gollhofer
Sept. 1994 – July 2003	Secondary school in Freiburg, qualification: A-levels
Sept. 1990 – July1994	Primary school in Freiburg

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APPENDIX

Table A1: Composition of patient groups.
The table lists the composition of the seven patient groups. The abbreviation for the patient groups are orthopaedic uni-/bilateral (OUni/OBi), neurologic flaccid uni-/bilateral (NflaUni/NflaBi), neurologic spastic uni-/bilateral with/without adequate trunk control (NspUni/NspBi/NspBiNTC).

Patient group	Diagnosis	Absolute Nr.	% of patients in the group
OUni	Patella dislocation	14	15.1
	Clubfoot	13	14.0
	Other knee problem (e.g. fractures, total endoprothesis, tumors, pain, instability)	12	12.9
	Knee ligament instability	9	9.7
	Other ankle problem (e.g. fractures, tumors)	8	8.6
	Torsional abnormality	6	6.5
	Developmental dysplasia of hip (DDH)	5	5.4
	Talipes equinus (pes eqinus)	3	3.2
	Malalignement of knee axis	3	3.2
	Perthes disease	3	3.2
	Hip pain/ instability	3	3.2
	Talipes calcaneus	2	2.2
	Planovalgus foot (pes planovalgus)	2	2.2
	Femoral deformity	2	2.2
	Other hip problem (e.g. fractures, total endoprothesis, tumors)	2	2.2
	Flatfoot (pes planus)	1	1.1
	Epiphyseolysis capitis femoris (ECF)	1	1.1
	General disease with unilateral problem (e.g. multiple osteochon dromas, achondroplasia, pseudohypoparathyreoidism, dysmorphic syndrome,TAR-syndrome, Turner-syndrome)	3	3.3
	Foot instability/pain/arthrosis	1	1.1
OBi	Leg length discrepancy	46	26.1
	Torsional abnormality	23	13.1
	Talipes equinus (pes eqinus)	20	11.4
	Clubfoot	15	8.5
	Patella dislocation	11	6.3

OBi	General disease with bilateral problem (e.g. multiple osteochondromas, achondroplasia, pseudohypoparathyreoidism, dysmorphic syndrome,TAR-syndrome, Turner-syndrome)	9	5.1
	Back pain	9	5.1
	Planovalgus foot (pes planovalgus)	7	4.0
	Arthrogyposis multiplex congenita	6	3.4
	Scoliosis	4	2.3
	Talipes calcaneus	3	1.7
	Flatfoot (pes planus)	3	1.7
	Malalignement of knee axis	6	3.4
	Hip (pain/ instability)	3	1.7
	Perthes disease, epiphyseal dysplasia	2	1.1
	Developmental dysplasia of hip (DDH)	2	1.1
	Foot instability/pain/arthroses	2	1.1
	Other knee problem (pain/ instability)	2	1.1
	Talipes valgus (pes valgus)	1	0.6
	Other ankle problem (e.g. fractures, tumors)	1	0.6
	Other spinal deformity (not scoliosis)	1	0.6
NflaUni	Nerve palsy (lower body)	8	66.7
	Poliomyelitis	4	33.3
NflaBi	Other neuromuscular diseases (e.g. Becker dystrophy, multiple sclerosis, myotonia, myopathy, HSMN, Polineuropathy, Myoclonic dystrophy (Curschmann-Steinert)	22	26.5
	Spina bifida	21	25.3
	Paraplegia	19	22.9
	Developmental retardation / coordination disorder / unclear brain disorder	12	14.5
	Duchenne's muscular dystrophy	5	6.0
	Down syndrome	2	2.4
	Spinal paralysis	1	1.2
	Poliomyelitis	1	1.2
NspUni	Hemiplegia (CP)	164	93.2
	Hemiplegia (not CP)	12	6.8
NspBi	Diplegia (CP)	116	97.5
	Spastic hereditary paraparesis	3	2.5
NspBiNTC	Tetraparesis (CP)	54	94.7
	Tetraparesis (not CP)	3	5.3