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Effects of idiopathic orthopedic deformities on gait biomechanics in children

Sara Kimberly Byrnes

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Summary

Background: Various orthopedic deformities of the lower limbs have been shown to affect gait biomechanics. Notably, the knee adduction moment (KAM) can be derived from three-dimensional gait analysis. KAM has received special focus, as it is a surrogate measure of the load distribution between the medial and lateral compartment at the knee. It is still unclear, however, as to whether compensatory mechanisms are used to counterbalance internal rotational malalignment, if these have an effect on KAM and if subcomponents of flatfoot deformity in children and adolescents correlate with KAM.

Research questions: The objective of this thesis was to perform a systematic review (study 1) on the factors influencing KAM during gait analysis and to, retrospectively, determine whether children and adolescents with internal rotational deformity of the lower limb show compensatory mechanisms to offset in-toeing, and whether these compensations affected KAM (study 2). It also examined whether hindfoot valgus, lateral calcaneal shift, forefoot abduction and lower medial longitudinal arch correlated with the magnitude of KAM in children and adolescents during walking (study 3).

Methods: A systematic review of the literature was performed according to the PRISMA checklist. Two databases, PubMed and Web of Science, were searched for peer-reviewed original research articles in which KAM was calculated during barefoot, three-dimensional gait analysis (study 1). In addition, two retrospective studies involving children and adolescents with internal rotational lower limb (n=69; study 2) and flatfoot deformity (n=103; study 3) and who had undergone standardized three-dimensional gait analysis were undertaken. Two-factor ANOVAs were performed to evaluate differences between children with and without internal rotational deformities. Independent and paired t-tests were used to evaluate differences in KAM in children with and without flatfoot deformity and following surgical correction. Pearson's correlations were used to explore potential relationships between KAM and flatfoot deformity.

Results: The systematic review included 42 studies and identified both gait-related and personal factors likely influence KAM. Gait-related factors included gait speed, lateral trunk lean, and step width, while personal factors included age, body weight and femoral or tibial torsions and genu varum/valgum. Cross-sectional analysis of children with idiopathic, internal rotational, orthopedic deformity of the lower limb (hip (n=25), tibia (n=18) or both (n=26)) showed that gait compensatory mechanisms were dependent on the location of the deformity (hip, tibia or both). For instance, children with internal tibial torsion had significantly greater external hip rotation (0.27° ± 7.19°) than typically developing children (5.65° ± 7.24°) during walking. KAM during walking, however, was comparable to that of typically developing children (TD). In distinct contrast, KAM in children with flatfoot deformity (0.37 Nm/kg ± 0.15 Nm/kg) was significantly reduced compared to that of TD children (0.54 Nm/kg ± 0.18 Nm/kg; P<.001). A moderate negative relationship was observed between KAM and lateral calcaneal shift (r=0.42), with lower KAM values associated with greater lateral calcaneal shift (P<.001).

Discussion: Previous research has associated increased KAM values with potential development of degenerative joint disease and/or pain of the knee. A systematic review of the literature identified that increased KAM was typically associated with fast walking speeds, a narrow step width, a toe-out gait pattern, femoral anteversion, external tibial torsion and genu varum. A number of compensatory mechanisms were found in children with internal rotational deformity of the lower limb, which likely acted to normalize KAM values to those of TD children. When considering children with flatfoot deformity during walking, evaluation of a newly introduced parameter, lateral calcaneal shift, may be clinically insightful, as it was the only subcomponent of flatfoot deformity that was associated with KAM.

Zusammenfassung

Hintergrund: Verschiedene orthopädische Fehlstellungen haben gezeigt, dass sie unterschiedliche Gangparameter beeinflussen. Das frontale Kniemoment (KAM), welches mithilfe von dreidimensionaler Ganganalyse erfasst werden kann, wird häufig in Untersuchungen als Indikator für die Belastungsverteilung zwischen dem lateralen und medialen Kniegelenkkompartiment herangezogen. Unklar ist jedoch, wie sich Kompensationsmechanismen zur Entgegenwirkung des Einwärtsgangs bei Innenrotationsfehlstellungen der unteren Extremität auf KAM auswirken und ob Subkomponenten des Knicksenkfußes mit KAM korrelieren.

Fragestellung: Eine systematische Literaturrecherche soll eine Zusammenfassung über die Faktoren der Ganganalyse liefern, die das KAM beeinflussen (Studie 1). Darüber hinaus wurden Gangdaten retrospektiv mit der Fragestellung untersucht, ob Kinder und Jugendliche mit Innenrotationsfehlstellungen der Hüfte und Tibia Kompensationsmechanismen nutzen, um ihr Einwärtsgehen auszugleichen und ob diese einen Einfluss auf KAM zeigen (Studie 2). Des weiteren wurde erfasst, ob es einen Zusammenhang von KAM und die Rückfußeversion, laterale Kalkaneusverschiebung und Vorfußabduktion sowie die Senkung der Fußlängsachse bei Kindern und Jugendlichen mit Knicksenkfuß während des Gehens gibt (Studie 3).

Methoden: Basierend auf der PRISMA Checkliste wurden für die systematische Literaturrecherche zwei Datenbanken, PubMed und Web of Science, durchsucht. Publikationen mit Untersuchungen des KAM während barfüßiger dreidimensionaler Ganganalyse wurden in die Analyse eingeschlossen (Studie 1). Für die retrospektive Datenanalyse ergab die klinische Datenbank 69 Kinder und Jugendliche mit Innenrotationsfehlstellungen (Studie 2) und 103 Kinder mit Knicksenkfuß (Studie 3), die eine standardisierte dreidimensionale Ganganalyse absolvierten. Eine zweifaktorielle ANOVA wurde durchgeführt, um die Unterschiede in der Kinematik und Kinetik zwischen Kindern mit und ohne Innenrotationsfehlstellungen zu analysieren. Mithilfe von t-tests wurden eventuelle Unterschiede von KAM zwischen Kindern mit und ohne Knicksenkfuß und nach korrigierender Operation evaluiert. Pearson’s Korrelationsanalysen untersuchten den Zusammenhang zwischen KAM und den Subkomponenten des Knicksenkfußes.

Ergebnisse: Die Ergebnisse der 42 eingeschlossenen Publikationen zeigten, dass besonderes Gehgeschwindigkeit, laterale Seitneigung des Oberkörpers, Schrittbreite, sowie Körpergewicht und Alter und femorale oder tibiale Torsionen und Genu Varum/Valgum eine Veränderung vom KAM hervorrufen. Die retrospektive Datenanalysen ergab, dass Kinder und Jugendliche mit Innenrotationsfehlstellungen bestimmte Kompensationsmechanismen abhängig von der Lokalisierung der Fehlstellung (Hüfte (n=25), Tibia (n=18) oder Beides (n=26)) aufweisen. Zum Beispiel, zeigte die Gruppe mit Tibiainnentorsion eine vermehrte externe Hüftrotation (0.27° ± 7.19°), um dem Einwärtsgang entgegen zu wirken im Vergleich zu orthopädisch unauffälligen Kindern (5.65° ± 7.24°). Das KAM zeigte jedoch ähnliche Werte zwischen den Gruppen. Bei Kindern mit Knicksenkfuß hingegen war das KAM deutlich niedriger (0.37 Nm/kg ± 0.15 Nm/kg) als bei orthopädisch unauffälligen Kindern (0.54 Nm/kg ± 0.18 Nm/kg; P<.001). Einen moderaten Zusammenhang gab es zwischen der lateralen Kalkaneusverschiebung und KAM (r=0.42). Je größer die laterale Kalkeneusverschiebung war, desto niedriger war das KAM (P<.001).

Diskussion: In vergangenen Studien konnte ein erhöhtes KAM mit potentieller Entwicklung von degenerativen Kniegelenkserkrankungen und/oder Schmerzen assoziiert werden. Folgende ungünstige Bedingungen, die das KAM erhöhen, sind basierend auf der Literaturrecherche gefunden worden: schnelles Gehen, schmale Schrittbreite, Auswärtsgang, Femorale Anteversion, externe Tibiatorsion und Genu Varum. Widersprüchliche Ergebnisse aus den Publikationen der Literaturrecherche führten zu der Untersuchung von Innenrotationsfehlstellungen. Bestimmte Kompensationsmechanismen konnten gefunden werden, die wiederum das KAM normalisieren könnten. Für die Evaluation des Knicksenkfußes sollte der neu beschriebene Parameter laterale Kalkaneusverschiebung in die Auswertung einer Ganganalyse aufgenommen werden, da dies die einzige Komponente des Knicksenkfußes war, die mit KAM einen Zusammenhang zeigte.

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Contents

Abbreviations

FPA .... *foot progression angle*
ITB .... *both, internal tibial and femoral torsion*
ITF .... *internal femoral torsion*
ITT .... *internal tibial torsion*
KAM .... *knee adduction moment, frontal plane knee moment*
KAM1 .... *first peak knee adduction moment*
KAM2 .... *second peak knee adduction moment*
KOA .... *knee osteoarthritis*
LCA .... *lateral calcaneus*
LMA .... *lateral malleolus*
MMA .... *medial malleolus*
OFM .... *Oxford foot model*
PRISMA .... *Preferred Reporting Items for Systematic reviews and Meta-Analyses*
STL .... *sustentaculum tali*

1. Background

In recent years, clinical gait analysis has become more popular in the field of pediatric orthopedics. By evaluating gait, specific treatment options, either conservative or surgical, can be selected and the outcome of the applied intervention can be assessed (Feng et al., 2016). Gait analysis is considered a meaningful tool for many conditions (Feng et al., 2016). This thesis will focus on two idiopathic orthopedic deformities of the lower limb that occur often in children and adolescents, namely idiopathic flatfoot deformity and internal rotational malalignment (Morley, 1957; Staheli et al., 1985).

1.1. Idiopathic orthopedic deformities

The term *idiopathic* indicates that the cause for the deformity is obscure or unknown and that the deformity often arises spontaneously (Merriam-Webster Dictionary); suggesting that there are no apparent underlying neurological or structural causes (Frances & Feldman, 2015). Focusing on the lower extremities, there are rotational, angular and foot deformities (Feng et al., 2016). However, first, "normal" limb alignment has to be defined, so that deviations can be readily identified and assessed. Lower extremity alignment changes during growth and development from the newly walking toddler to the fully grown adolescent. It is therefore greatly age dependent (Espandar et al., 2010; Scorcelletti et al., 2020; Staheli et al., 1987). However, some malalignments persist outside of the range of typical alignment variations and must be observed and checked regularly, since these could directly or indirectly induce pain, overuse injuries or degenerative diseases in the future (Baliunas et al., 2002; Bramah et al., 2018; Sharma et al., 2010; Turner, 1994). Deformities sometimes resolve with growth or with support of conservative treatment (e.g., insoles, orthotics, physiotherapy) (Frances & Feldman, 2015; Staheli et al., 1985). However, for some malalignments, surgical intervention is considered more appropriate during childhood and adolescence, as "growth" provides a correction opportunity (e.g., "guided growth" by hemiepiphysiodesis in patients with knee valgus/varus alignment (Eastwood & Sanghrajka, 2011)). However, flatfoot deformity and internal rotational malalignment in children are usually only surgically managed if persisting deformity and symptoms are present (Davids et al., 2014; Frances & Feldman, 2015). Hence, knowledge concerning the nature of the deformity, its characteristics and progression should aid pediatric orthopedic surgeons to act reasonably in treatment and intervention planning. Three-dimensional instrumented gait analysis is one approach that can be used to objectively identify the impact of deformity on other body structures and functions during daily activities of living. While three-dimensional gait analysis will be reviewed in more detail in Section 1.2.1, it is first necessary to discuss the specific deformities that are evaluated in this thesis.

1.1.1. Lower extremity internal rotational malalignment

First, a clear definition of the terms rotation and torsion is necessary. While torsion describes the twist in the axis of the same segment (i.e. femur, tibia), rotation describes the movement of the segment around an axis (Rosen & Sandick, 1955). The term torsion is therefore used when the malalignment itself is referred to, such as femoral/tibial torsion, whereas rotation is further used when reporting on movements, such as gait kinematics (e.g., hip rotation).

The bony structures involved in the alignment of the leg axis are the femur, patella, tibia and fibula (see Figure 1). The relative axial torsion of the two segments of the lower extremity, femoral anteversion and tibial torsion, physiologically change during growth (Staheli et al., 1985). Physiological passive internal and external rotation of the hip is around 40-50° in each direction, when measured clinically with the child in a prone position and flexed knees. For the tibia, the angle of transmalleolar axis increases during the first few years of life and is approximately 25° externally rotated relative to the thigh during puberty; decreasing slightly to

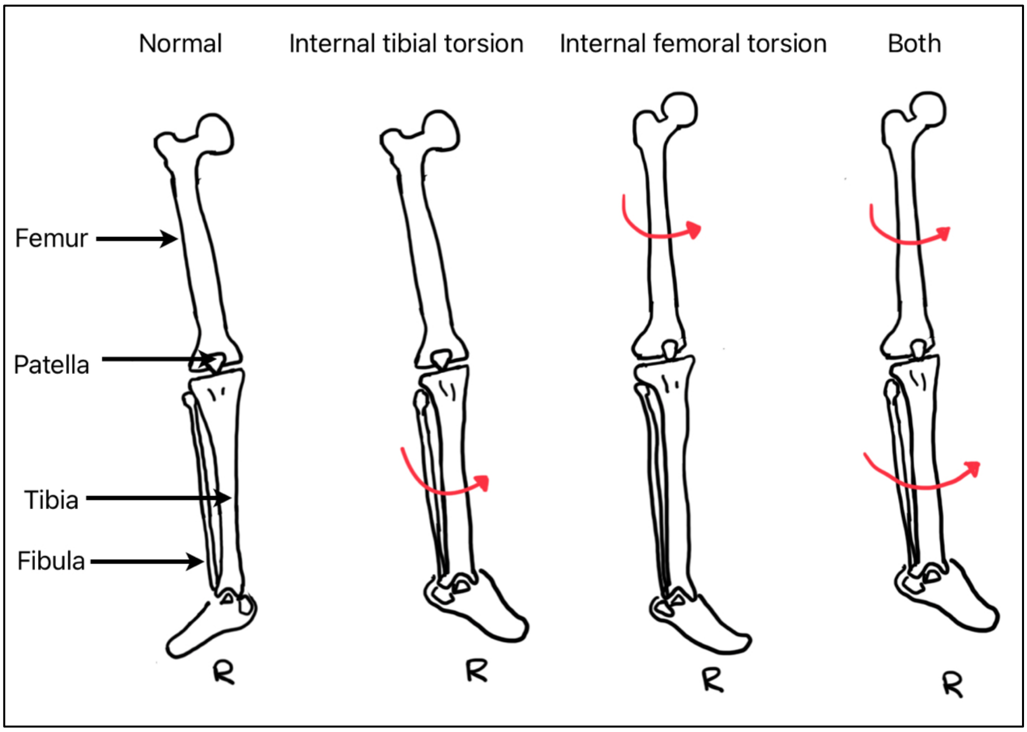

Figure 1. Visual depiction of the internal torsional deformities. Depicted is the right (R) leg for A) normal torsion B) internal tibial torsion C) internal femoral torsion and D) both, femoral and tibial torsions combined.

around 20° in adulthood. Since the present thesis investigates the effect of overall increased internal torsions on gait biomechanics, this section will focus on internal torsions of the femur and decreased external torsion of the tibia. In this thesis, decreased external torsion of the tibia is referred to as internal tibial torsion, since an angle below 0° (therefore a "true" internal torsion) is rare (Staheli et al., 1985).

Internal torsion of the lower extremity can dominate and persist during growth and development instead of decreasing with age. Internal rotational malalignment of the lower extremity is easily diagnosed by clinical examination, while internal torsion of the bones is diagnosed by computer tomography. The femur or tibia can be affected in isolation or in combination (Lerch et al., 2019; Staheli et al., 1985). In clinical practice, surgical treatment may be indicated for symptomatic malalignment and marked deviation from normal values (Davids et al., 2014). To obtain the intended outcome, it is important to consider the compensatory gait deviations that may occur (see section 3.2) and follow up on these post-surgery by three-dimensional instrumented gait analysis, as these should disappear with the correction of the malalignment. It has been reported that compensatory mechanisms remain after surgery, which could lead to further problems (Davids et al., 2014). Therefore, surgical treatment needs to be well reasoned.

1.1.2. Flatfoot deformity

Pes planovalgus or more commonly termed *flatfoot deformity* is a foot deformity that is characterized by an under developed medial longitudinal arch, resulting in a "lowered" or even completely flat foot, as seen clinically (Mosca, 2010). The talo-navicular joint becomes decentered, which may further lead to a subluxation of the subtalar joint (Lashkouski, 2007). Furthermore, flatfoot deformity typically involves a multi-dimensional combination of

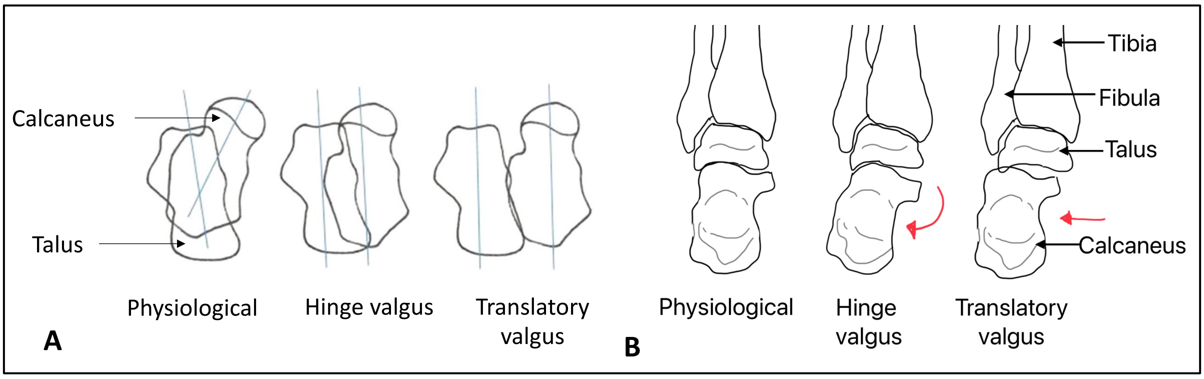

Figure 2. Illustration of the difference in rotatory and translatory hindfoot valgus as viewed in a dorsoplantar (A) and posterior-anterior (B) direction.

deviations, often affecting more than just one plane to involve all three planes (sagittal, frontal and transversal) (Bourdet et al., 2013; Mosca, 2010). The following subcomponents may dominate in flatfeet: a low medial longitudinal arch height (sagittal plane), hindfoot valgus (frontal plane), and forefoot abduction (transversal plane), and supination. The subcomponent, hindfoot valgus, may further occur secondary to eversion of the hindfoot (hinge valgus) or to a lateral shift of the hindfoot (translatory valgus) or both (see Figure 2) (Eberhardt et al., 2018; Thompson & Abaza, 2010). Flatfoot is a common variant during child development, being highly prevalent in early childhood and significantly decreasing with age. Flatfoot deformity was found in 97 % of 1.5 year-old children, but in 54 % of 3 year-old children and in only 4 % of 10-11 year-old children (Morley, 1957; Pfeiffer et al., 2006). For the deformity to be considered *idiopathic*, the examining orthopedic pediatrician should rule out other causes that can lead to flatfoot deformity, such as structural changes (e.g., tarsal coalition), neuromuscular diseases (e.g., muscle dystrophies, cerebral palsy) and syndromes (Frances & Feldman, 2015). However, a clear and standardized definition for the diagnosis of flatfoot deformity is currently lacking due to the absence of precise differentiation between normal and pathological variations of the foot shape (Mosca, 2010). Until now, orthopedic pediatricians subjectively diagnosed flatfoot deformity based on the collapse of the medial longitudinal arch, the valgus of the hindfoot and the tightness of the Achilles tendon (Frances & Feldman, 2015), in addition to precise and detailed clinical and radiographical examination and under consideration of differential diagnoses. Typically, idiopathic flatfoot deformity is painless with no indication for specific treatment (Frances & Feldman, 2015). However, in symptomatic children or adults with flatfoot deformity, conservative treatment may help ease the pain and symptoms (Frances & Feldman, 2015). If pain persists or becomes unbearable, surgical treatment might be indicated to correct the deformity (Frances & Feldman, 2015). Moreover, flatfoot deformity is reported to be associated with pain of other body regions external to the foot, such as the knee, hip or back (Hösl et al., 2014; Kothari et al., 2016). Hence, it is important to perform a holistic examination of the patient presenting with pain. Children with persistent deformity and who participated in the following study, with or without existent pain, were referred to the gait analysis laboratory at the Children's Treatment Center in Aschau i. Chiemgau, Germany, where further treatment and therapy can be planned based on additional examination via gait analysis.

1.2. Gait biomechanics

1.2.1. Three-dimensional instrumented gait analysis

To obtain specific biomechanical gait parameters, three-dimensional instrumented gait analysis is performed. This method is especially useful for the non-invasive investigation of the joint and body segment movement, to gain insight into joint loading, and to support clinical decision-making (Stief, Holder, Böhm, et al., 2021). For many children, where clinical examination may not seem sufficient to establish a clear diagnosis or treatment plan, referral to a specially-equipped, gait laboratory is becoming increasingly common.

Both retrospective studies undertaken in the present thesis (study 2 and study 3) were based on such clinical data; extracted from three-dimensional instrumented gait analysis (see Figure 3) performed within the last 10 years at the gait laboratory at the Children's Treatment Center in Aschau im Chiemgau, Germany. The procedure of clinical gait analysis in the Center is well established and standardized and follows a specific pattern, which allows for the conduct of research. The following description of methods and tools used is, therefore, similar for both retrospective studies and is a repetition to the methods section presented in each publication (see Appendix).

An eight-camera (200 Hz) motion analysis system (VICON Motion Systems Ltd., Oxford, UK) and two standard force platforms (1000 Hz, AMTI, Watertown, MA, USA) imbedded in an approx. 13-meter walkway was used to record children while walking at their own self-selected speed. Prior to data collection, children were prepared and equipped with reflective markers, which for reliable measurement, needed to be placed by trained personnel on specific bodily landmarks for reliable measurement. For the first retrospective analysis, these were placed according to a modified Plug-In-Gait marker set (Stief et al., 2013), where medial ankle and knee markers were added during the static measurement to allow for a more accurate calculation of joint rotations in the transverse plane, which is especially relevant for the investigation of lower extremity rotational malalignments.

The elaborate, non-invasive analysis allows for calculation of spatio-temporal parameters, such as step width, step length, and gait velocity, along with collection of kinematic and kinetic data. This was done by taking the three-dimensional marker positions in space and calculating the body segment and joint movements (kinematics) and by using ground reaction force data gained from force platforms to estimate joint moments and joint contact forces using inverse dynamics methods (kinetics) (Davis et al., 1991; Stief, Holder, Böhm, et al., 2021). Therefore, it was possible to analyze the gait in all three body planes (sagittal, frontal and transverse; see Figure

3) and to extract outcome variables for all joints (pelvis, hip, knee, ankle and foot) for the movements of flexion and extension (sagittal plane), abduction and adduction (frontal plane) and internal and external rotation (transverse plane).

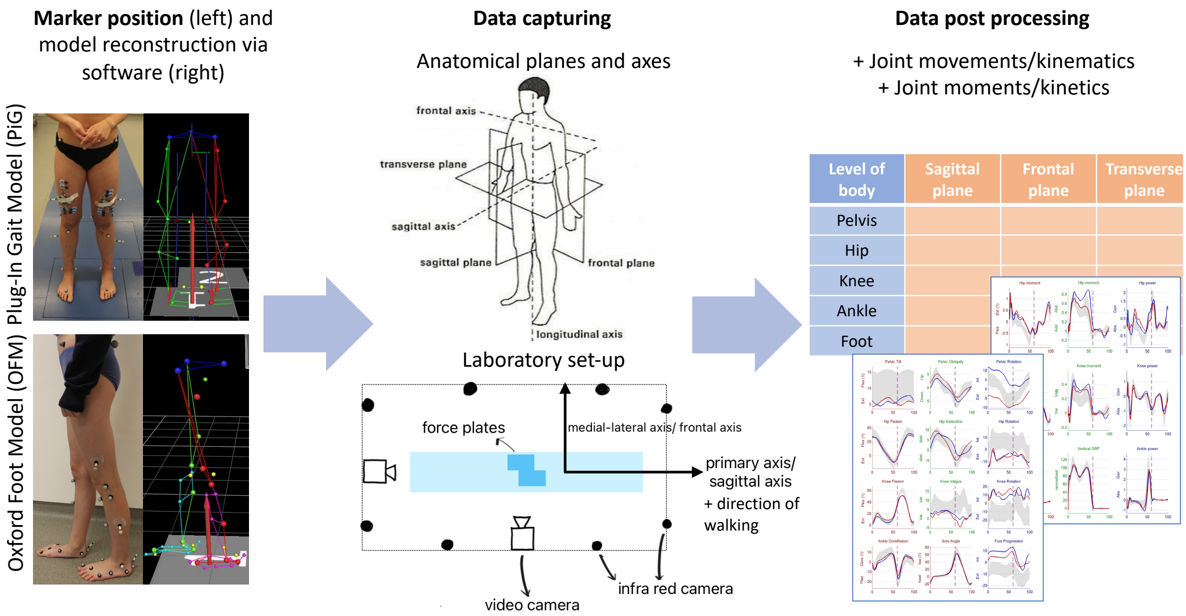

Figure 3. Three-dimensional gait analysis procedure. 1. marker position on specific body landmarks according to marker model, either Plug-In Gait model or Oxford foot model. 2. gait data capturing in gait laboratory via force plates imbedded in the walkway and infrared cameras installed at the ceiling around the walkway; make it possible to evaluate all three movement planes (figure by (de Oliveira Sato et al., 2010)) 3. Results of three-dimensional gait analysis: joint kinematics and kinetics for lower body joints for all three planes.

To obtain gait kinetics, the ground reaction force vector (see Figure 5) recorded by the force plates during walking is taken into the calculations via the inverse dynamics method (Davis et al., 1991). The area of the sole where the force is acting upon is the center of pressure (Richards, 2008) or the point of force application (Chockalingam et al., 2016). In more detail, the point of force application basically moves along the foot during walking as the foot rolls over the ground

Figure 4. Pressure distribution with physiological foot shape during walking. Illustrating the areas under the foot where the ground reaction force vector applies (black line).

(Jamshidi et al., 2010). Physiologically, starting at the heel during initial contact, moving along the midfoot and ending at the forefoot during push-off phase (see Figure 4 and Figure 5). The idea is that depending the position of the foot structures (i.e. hindfoot, midfoot, forefoot) in relation to the ground reaction force vector the outcome of gait kinetics may differ.

The knee adduction moment (frontal plane knee joint moment or KAM) is one of the kinetic outcome variables of three-dimensional instrumented gait analysis and is known to be influenced by several gait or body-related (e.g. foot) factors (see study 1). Over the course of the stance phase of gait, the KAM typically shows two distinctive peaks. The first peak (KAM1) occurs during loading response and the second (KAM2) during terminal stance phase of gait (Figure 5). As described above, physiologically the hindfoot has the first ground contact during walking, where KAM1 occurs and the forefoot pushes off the ground where KAM2 occurs (see Figure 5). Hence, it is possible that deformities concerning the hindfoot may specifically impact KAM1, while deformities concerning the forefoot may selectively impact KAM2.

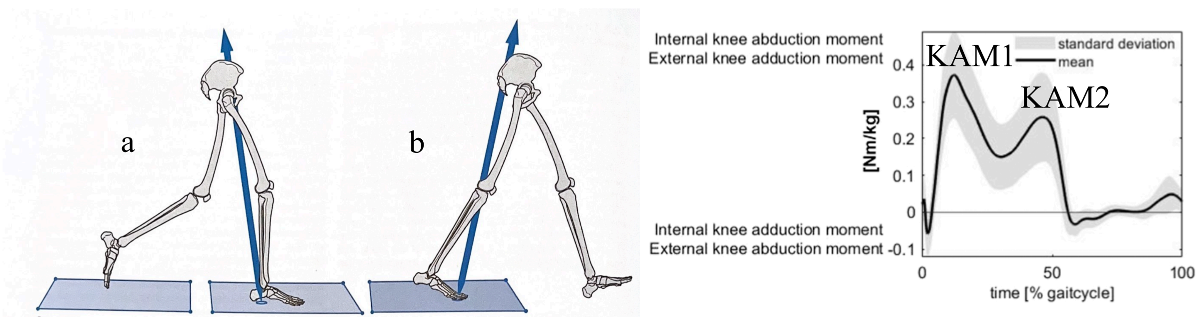

Figure 5. Left: a) ground reaction force vector during loading response phase and b) ground reaction force vector during push-off phase (Richards, 2008, p. 40). Right: A general illustration of pseudo data of the knee adduction moment (figure taken from study 1). Peaks of KAM represent higher forces acting on the joints during loading response (KAM1) and push-off phase (KAM2).

KAM has been a widely and intensively researched and relevant parameter in both, research and clinical settings, as a surrogate measure of the load distribution between the medial and lateral compartment of the knee joint in the frontal plane (Birmingham et al., 2007; Zhao et al., 2007). KAM1 has also been associated with knee osteoarthritis (KOA) (Baliunas et al., 2002). Hence, depending on structural malalignments or dynamic changes (gait deviations), the load within the knee joint may shift more towards the lateral or medial compartment, causing an imbalance of load distribution which may, in turn, induce musculoskeletal pain (Amin et al., 2004), and cartilage degeneration over time (Andriacchi & Mündermann, 2006; Baliunas et al., 2002; Mündermann et al., 2005; Sharma et al., 1998). As depicted in Figure 6, the load can shift towards the lateral side for a child with flatfoot deformity (left blue box), probably due to the lateralization of the heel (hindfoot valgus) resulting in a lower KAM compared to children with a normal shaped foot (right black box in Figure 6).

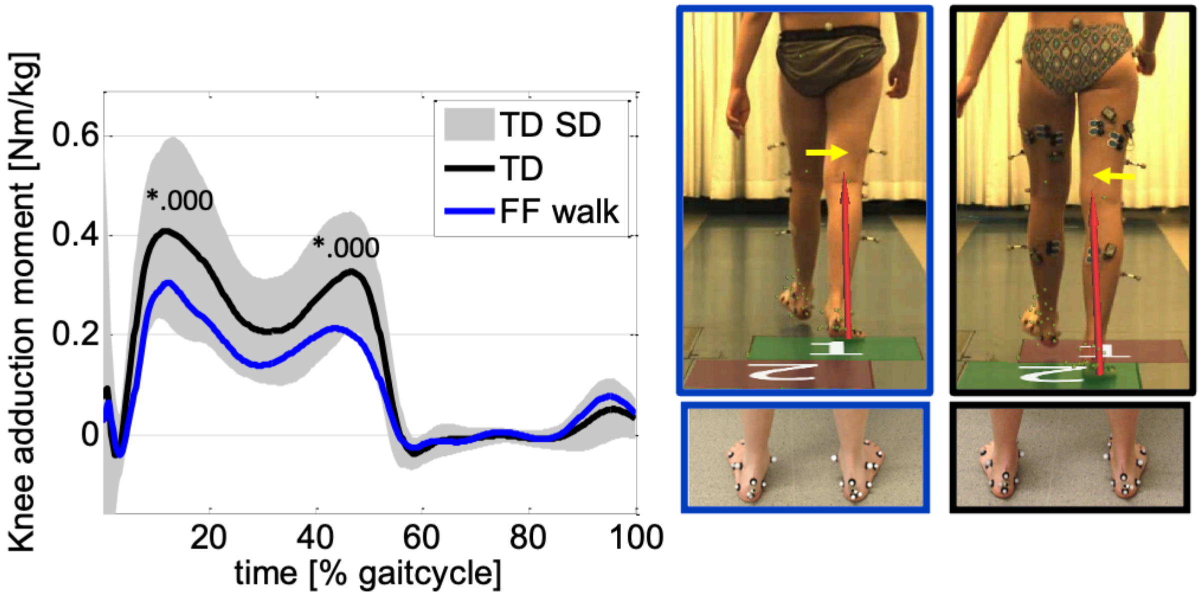

Figure 6. Left: KAM is depicted for children with (blue, FF) and without (black, TD) flatfoot deformity. Both peaks are significantly reduced in children with flatfoot deformity. Right: Illustration of the projection of the ground reaction force vector in relation to the frontal plane knee joint center in a child with (FF) and without (TD) a flatfoot deformity. The blue box (left) shows a child with flatfoot deformity in which the force vector is directed more laterally, than that of a child without orthopedic malalignment (black box, right), where the force vector passes physiologically to be slightly medial of the knee joint center.

Oxford foot model marker set

For the second retrospective analysis, in addition to the standard Plug-In-Gait marker set, additional foot markers were placed according to the Oxford foot model (OFM) (Stebbins et al., 2006). The set-up allowed a detailed investigation of the subcomponents of flatfoot deformity (i.e. hindfoot eversion, low medial longitudinal arch, forefoot abduction and supination) in relation to KAM. However, the standard outcome parameters of this model do not allow for the differentiation between rotatory and translatory hindfoot valgus (Thompson & Abaza, 2010). Therefore, this thesis introduced, for the first time, the lateral calcaneal shift parameter, which could be calculated based on the OFM marker set (study 3). The midpoint between the three-dimensional coordinates of the medial (MMA) and lateral (LMA) ankle markers was projected onto the line between medial (STL) and lateral (LCA) heel markers. The distance from the projected point to the midpoint in percentage (to account for heel size differences and comparability) of the calcaneal width (distance LCA-STL) represents the measure lateral calcaneal shift (see Figure 7).

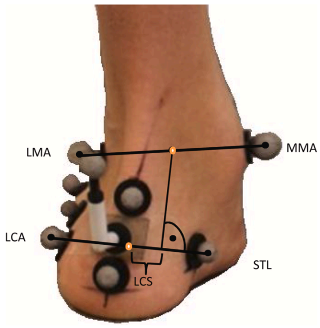

Figure 7. Illustration of how the lateral calcaneal shift (LCS) is calculated. MMA: medial malleolus. LMA: lateral malleolus. LCA: lateral calcaneus. STL: sustentaculum tali (taken from study 3).

The next section provides a detailed overview as to how internal rotational malalignment of the lower extremity and flatfoot deformity reportedly affect the gait of children as reported by previous research.

1.2.2. Effect of internal rotational malalignment on gait biomechanics

Internal rotational malalignment can result in obvious (changed FPA) or not immediately apparent (compensatory mechanisms) changes in gait biomechanics. For instance, increased hip rotation (internal rotation) and FPA during walking may be present as an obvious gait change to an internal femoral and tibial torsion deformity, resulting in in-toeing walking (Radler et al., 2010). Most parents typically observe abnormal in-toeing walking in their children, rather than the deformity itself, and subsequently present to orthopedic specialists. Oftentimes, children and parents report an increased frequency of tripping and falling incidents as a result of in-toeing walking (Davids et al., 2014; Leblebici et al., 2019), as the internally rotated swing leg collides with the stance limb or the toes do not gain enough clearance from the ground, when swinging the leg forward. However, not all children with internal rotational malalignment present with in-toeing walking (Fabry et al., 1994; Thackeray & Beeson, 1996). For instance, children with internal femoral torsion are thought to develop a compensatory external tibial torsion (i.e. miserable malalignment) to offset in-toeing walking; thereby presenting with a normal FPA (Fabry, 1977; Radler et al., 2010). Furthermore, children may develop compensatory mechanisms or gait strategies to counterbalance the increased FPA (internal foot rotation) to avoid tripping and falling. For example, it was shown that children with internal tibial torsion actively externally rotate their hip during walking, in an attempt, to normalize their FPA (Davids et al., 2014). In contrast, children with internal femoral torsion may present with greater pelvic retraction (external rotation) (Radler et al., 2010), anterior pelvic tilt, knee adduction and hip flexion (Bruderer-Hofstetter et al., 2015). Previous research has shown that internal femoral and tibial torsion influence KAM (Davids et al., 2014; MacWilliams et al., 2016), and should, therefore, be considered by clinicians when treating patients with internal rotational malalignment. These previous studies have focused on one level of deformity (femur or tibia). In this thesis the aim was to evaluate compensatory mechanisms in children with either internal femoral torsion, internal tibial torsion or a combined deformity of both, internal femoral and tibial torsion and how these affect the outcome of KAM during walking at preferred speeds.

1.2.3. Effect of flatfoot on gait biomechanics

Previous research has shown that flatfoot deformity may not only effect the kinematics of the foot during gait (Saraswat et al., 2014), but may also impact kinematics and kinetics at different joint levels (Kothari et al., 2016). For instance, Kothari et al. (2016) observed that flatfoot deformity in children increased pelvic retraction (external rotation) and knee valgus during walking, which may be the reason for symptomatic children with flatfeet reporting pain in more proximal joints, not only at the foot level. Interestingly, Kothari et al. (2016) also observed that the KAM was reduced in children with flatfoot deformity compared to children with a neutral foot posture (Kothari et al., 2016). While the reduction in KAM1 approached statistical significance, KAM2 was markedly and significantly reduced. However, the authors did not report on the effects of the different subcomponents of flatfoot deformity on KAM. Therefore, this thesis aimed to identify the effects of the flatfoot subcomponents (hindfoot eversion, lateral calcaneal shift, forefoot abduction) on KAM.

1.3. The research gap

In the literature, several variables have already been investigated to show their influence on KAM. These are important to consider, especially when clinical decisions are based on the outcome of pathological KAM. For example, in clinical practice, a reduction in gait velocity and medio-lateral trunk sway has been used as an intervention in KOA patients as a way to lower their medial knee joint loading (Mündermann et al., 2004, 2008). Different gait strategies or modifications and footwear have also been shown to influence KAM in healthy and osteoarthritic patient populations and have been summarized previously in two systematic reviews (Simic et al., 2011; Telfer et al., 2017). However, idiopathic orthopedic deformities also influence KAM (Davids et al., 2014; Farr et al., 2017; Kothari et al., 2016; MacWilliams et al., 2016), and if the deformities are marginal and go unnoticed, findings from the gait analysis could be misinterpreted. Furthermore, some idiopathic orthopedic deformities, such as increased external tibial torsion, may induce compensatory mechanisms, such as internal hip rotation (Alexander et al., 2020), which can be considered a kind of gait modification, and in turn may influence the KAM values. Therefore as a part of this thesis, a systematic review was undertaken to summarize the evidence and to classify factors related to the conduct of clinical gait analysis that influence KAM, including idiopathic orthopedic deformities, as an addition to the previously published reviews (Simic et al., 2011; Telfer et al., 2017). Furthermore, it was not clear whether children with internal rotational lower extremity malalignment, especially combined femoral and tibial torsion, present with compensatory mechanisms and if these affect KAM. Finally, although previous research has shown that flatfoot deformity tends to lower KAM, the influence of the subcomponents of flatfoot deformity on the two peaks of KAM has not yet been investigated.

2. Objectives

The overall aim of the present thesis was to broaden the scope of clinical research findings considering the parameter KAM in order to support future data interpretation, especially for internal rotational and flatfoot deformities and their therapy and treatment planning in pediatric orthopedics. A special focus of the research was to investigate the effects of idiopathic orthopedic deformities of the lower extremity that may lead to a changed KAM in children during walking. The first objective of this thesis (study 1) was to review and classify participant-related aspects during gait analysis that may influence KAM during gait analysis. This objective was addressed by a systematic review of the gait and biomechanics literature. Studies were included and summarized that investigated barefoot walking within an asymptomatic population, as well as in populations with idiopathic orthopedic deformities. To ensure broad coverage, children and adult populations were included within the systematic review. Although children are generally not subject to osteoarthritis, the majority of published studies that have been conducted to date have focused on adults. The systematic review was published in the Journal of Gait & Posture with the title “Frontal plane knee moment in clinical gait analysis: A systematic review on the effect of kinematic gait changes” (Byrnes et. al. 2022, Appendix 7.1, page 32).

Furthermore, the impact of gait deviations on KAM produced by idiopathic orthopedic deformities was determined, e.g., compensatory mechanisms to offset primary deformity or just the deformity itself, e.g., internal torsional deformity. The second objective (study 2) was to explore whether children and adolescents with internal torsional lower extremity malalignment show compensatory mechanisms and if these impact the KAM. It was hypothesized that children and adolescents with internal torsional malalignment of the lower extremity would present with compensatory mechanisms, such as external hip rotation, increased knee, hip and ankle flexion and greater step width, to offset in-toeing during walking at preferred speed. Further, it was proposed that such compensatory strategies, while normalizing FPA, might also act to influence KAM. The results of this study titled "Compensatory mechanisms in children with idiopathic lower extremity internal rotational malalignment during walking and running" were published in the Journal of Gait & Posture (Byrnes et. al., 2020, Appendix 7.2, page 43).

Finally, the third objective (study 3) was to investigate the effect of, and association among, the subcomponents of flatfoot deformity in children with the two peaks in KAM magnitude during gait. Based on basic biomechanical principals, it was hypothesized that the magnitude of hindfoot valgus and lateral calcaneal shift would be correlated with KAM1 (loading response phase), while the magnitude of forefoot abduction and medial longitudinal arch height would be correlated with KAM2 (push off phase) secondary to the relative trajectory of ground reaction forces to the knee joint center. The results of this study were published in the manuscript "Effects of idiopathic flatfoot deformity on knee adduction moments during walking" (Byrnes et. al., 2021, Appendix 7.3, page 51) in the Journal of Gait & Posture.

All measurements for the retrospective studies (study 2 and study 3) were performed within the last 10 years, and took place at the gait laboratory located in the Children's Treatment Center in Aschau im Chiemgau, Germany.

The following chapter provides an overview and summary of the key findings of each study.

3. Summary of included studies

3.1. Study 1: Frontal plane knee moment in clinical gait analysis: A systematic review on the effect of kinematic gait changes

The frontal plane knee moment or KAM has been widely used as a surrogate measure of the load distribution between the medial and lateral compartment of the knee. Therefore, it has been of great interest in research as well as in clinical settings. The aim of this systematic review was to generate an overview of the factors influencing KAM, as they are important to consider when conducting a research study or interpreting data in clinical settings. On the basis of the PRISMA checklist, two databases, Pubmed and Web of Science, were screened for peer-reviewed, original research articles that investigated the KAM in children and adults during gait. In total, 42 studies were eligible for inclusion. Included studies were grouped into three categories, based on the independent variable investigated: 1) gait modifications, 2) individual characteristics and 3) idiopathic orthopedic deformities. Since KAM1 has been commonly associated with degenerative knee diseases and pain within the literature, only factors increasing the first peak are enumerated in this summary. An extensive list of factors can be found in the publication itself. In terms of gait modifications, a fast walking speed, narrow step width and out-toeing gait were identified to increase KAM1. Looking at individual characteristics, the dominant limb (gait initiating limb) and advancing age seem to show higher KAM1 during walking. Similarly, external tibial torsion was found to increase KAM1 (idiopathic orthopedic deformities). However, there were discrepancies in results reported within the literature for femoral anteversion and low arched feet. One study (Alexander et al., 2019) found an increase of KAM1 in individuals with femoral anteversion, while others report a decrease in KAM1 (MacWilliams et al., 2016). Considering the foot arch, one study showed that both low and high arches had no influence on KAM during walking (Buldt et al., 2015), whereas other studies reported lower KAM1 and KAM2 in individuals with low arched feet (Kothari et al., 2016) and also for high arched feet (Powell et al., 2016).

Illustration of the author's contribution

The PhD-candidate was responsible for the conceptualization and methodology of this review including the systematic search. She administered the project, designed the necessary MATLAB scripts for visualizing and summarizing the data, performed the formal analysis and drafted and revised the manuscript with assistance from the co-authors. As corresponding author, the PhD-candidate interacted with the journal during the submission and reviewing process.

3.2. Study 2: Compensatory mechanisms in children with idiopathic lower extremity internal rotational malalignment during walking and running

This study investigated the effect of in-toeing gait, secondary to torsional deformities of the lower extremities (femoral and/or tibial internal torsion), on gait kinematics in children during walking. In children, in-toeing often results from an internal torsional deformity of the lower leg and may lead to tripping and falling incidents. The aim of this retrospective data analysis was to determine if compensatory mechanisms are present in children with internal torsional malalignment of the lower extremity. And furthermore, if compensatory mechanisms exist, how they impact KAM. Gait data extracted from three-dimensional gait analysis was compared between children with isolated internal femoral torsion (ITF, n=25), with isolated internal tibial torsion (ITT, n=18) and with both malalignments (ITB, n=26) to age-matched typically developing children (TD, n=22). Compensatory mechanisms to reduce the effect of in-toeing were observed, but were dependent on the location of the torsional deformity. Pelvic retraction (external rotation) during walking was increased for the ITF and ITB groups (hip affected groups), which might contribute to improved toe clearance to aid in minimizing tripping and falling incidents. Although step width was greater for all torsional groups, only ITT showed greater external hip rotation and greater second peak KAM during walking. The clinical implication of a greater KAM2 remains unknown, however, it could still influence long term knee joint health or provoke the development of pain. KAM1, on the other hand, has previously been associated with degenerated knee joint health, but KAM1 values in this study were similar for all groups compared to TD children. Compensatory mechanisms may be the reason for these nearly normalized values and should further be observed especially when changes occur, such as corrective treatment of the malalignment.

Illustration of the author's contribution

The PhD-candidate was responsible for the conceptualization and methodology of this study. She searched the clinical database to gather the necessary data for retrospective analysis. She performed all statistical analyses, was responsible for data interpretation and visualization of the data, as well as drafting and revising the final manuscript with assistance from the co-authors. The PhD-candidate was responsible for the submission of the manuscript and drafted revisions secondary to the review process.

3.3. Study 3: Effects of idiopathic flatfoot deformity on knee adduction moments during walking

Idiopathic flatfoot deformity represents a combined deformity, with varying scope of a low medial longitudinal arch, hindfoot valgus and forefoot abduction and supination. Hindfoot valgus may, in turn, present as either a lateral shift of the heel, an eversion of the heel with respect to the leg or their combination. In this retrospective study, the individual effects of each specific subcomponent of idiopathic flatfoot deformity on KAM were investigated in children during walking at preferred speed. It was hypothesized that KAM1 would be lower in those with a lateralized hindfoot and KAM2 would be reduced during toe-off phase in those with abduction of the forefoot. Three-dimensional gait analysis data obtained from 103 children and adolescents with flatfoot deformity were analyzed and compared to the data of 15 TD children with a normal rectus foot type. Since the Oxford foot model (OFM) only includes hindfoot eversion and does not account for estimates of lateral shift of the calcaneus, a new parameter (lateral calcaneal shift) was introduced in this study and calculated based on OFM marker set. The results of this study demonstrated that both peaks KAM1 and KAM2 were reduced in children with flatfoot deformity compared to children with a rectus foot type. A weak to moderate linear relationship was observed between the lateral calcaneal shift and KAM1 and KAM2 was found ($r=.42$ $p<.001$ and $r=.32$ $p<.001$, respectively), with greater lateral shift associated with lower KAM values. Medial longitudinal arch height, in contrast, showed only a weak relationship with KAM2 ($r=.23$ $p<.001$). Standard measures of hindfoot eversion and forefoot abduction derived from the OFM were not significantly related to KAM. Although further research is needed, measurement of lateral calcaneal shift might therefore prove to be a more clinically relevant indicator of frontal plane alignment of the hindfoot in evaluations of flatfoot deformity in children and adolescents.

Illustration of the author's contribution

The PhD-candidate was responsible for the conceptualization and methodology of this study. She gathered all the data retrospectively from the clinical database, performed the data analysis, including the interpretation of statistical analyses, visualization and discussion. The original draft of the manuscript was written by the PhD-candidate, who was responsible for submitting the manuscript and overseeing the review process as corresponding author.

4. Discussion

In light of the present thesis, three studies have been published that collectively provide new insights into factors that influence measurement of KAM during instrumented three-dimensional gait analysis. It has also detailed the role of internal rotational malalignment of the lower extremity on gait compensations and of the individual components of flatfoot deformity and KAM in children and adolescents during walking.

The systematic review (study 1) provided an important summary to support orthopedic physicians and researchers in using and interpreting gait data. The review identified a number of participant-related factors that influence KAM, which were categorized into three groups according to the independent variables investigated: 1) gait modifications, 2) individual characteristics and 3) idiopathic orthopedic deformities. It is recognized, however, that these groups are not mutually exclusive but rather intermixed. For instance, gait can be intentionally modified to counterbalance pain or can be subconsciously changed to compensate for lower limb malalignments, such as flatfoot or internal rotational deformity of the tibia or femur. Intentional changes of gait were classified within the group of factors relating to "gait modifications", whereas changes associated with lower limb malalignments were summarized within the classification of "idiopathic orthopedic deformities".

Intentionally modified gait may deliberately reduce medial knee joint loading, which seems to be of interest in persons with knee pain or KOA (Simic et al., 2011). Preferred gait speeds are typically associated with minimal movement variability when compared to faster and slower speeds (Jordan et al., 2007), but can be highly variable and context dependent (Chang et al., 2018; Finley & Cody, 1970; Knoblauch et al., 1996). For instance there is evidence that measurements of gait speed in laboratory settings, an unfamiliar environment in which individuals are requested to walk "as normal as possible", may still differ to habitual walking when measured in more familiar urban environments (Corrà et al., 2021; Foucher et al., 2010; Krumpoch et al., 2021). Moreover, when trying to compare multiple measurements within a single person, control of gait speed is typically advocate to be within a 5% margin, to ensure consistency of data. This is particularly important, as the systematic review identified that gait speed reliably influenced the magnitude of KAM measures (Schwartz et al., 2008; van der Linden et al., 2002). Similarly, lateral trunk lean/sway was found to consistently lower KAM during gait (Anderson et al., 2018; Robbins et al., 2016; van den Noort et al., 2013). Hence, modification of gait speed and trunk sway might be one of the main clinical strategies used to modulate knee joint load and pain in specific clinical groups, as the literature on this topic seems to be in complete agreement. On systematically reviewing the literature, intentional in- and out-toeing has been reported to have mixed effects on KAM. Both in-toeing and out-toeing have been found to lower KAM. Whereas other studies have reported that in-toeing reduces only KAM1 (Cui et al., 2019; van den Noort et al., 2013), while out-toeing reportedly reduces KAM2 (Lynn et al., 2008; van den Noort et al., 2013). Therefore, combining specific gait modifications may be an individually successful management to reduce the overall knee moment, e.g., out-toeing plus wider step width (Stief, Holder, Feja, et al., 2021).

As further identified through the literature review (study 1), gait can also be subconsciously and subtly modified, not necessarily by underlying malalignments but by person-specific deviations, such as a decrease of thoracic kyphosis or an individual gait pattern (Kulmala et al., 2013; Ota et al., 2015). Subtle deviations might also be important to consider, when evaluating gait data, as they may not show obvious gait changes to the examiner. As a person-specific, individual characteristic, being overweight was found to reduce KAM, presumably due to a greater knee valgus angle in overweight populations (McMillan et al., 2009, 2010).

Underlying idiopathic orthopedic deformities with anatomical structural changes can either directly or indirectly (due to compensatory mechanisms) modify gait patterns. This was the focus of the second objective of this thesis. In study 2, children with internal rotational malalignment were divided into three groups, those solely with internal tibial torsion (ITT), those solely with internal femoral torsion (ITF) and children who present with both deformities (ITB). In-toeing gait was present in all groups and, in agreement with previous research (Radler et al., 2010), mostly involved the ITT group (100%); 92% in the ITB and 76% in the ITF group. Hence, some children with an internally rotated hip, around one quarter in this study, may not present with in-toeing gait. Children that had both an internally rotated hip and tibia (ITF and ITB), in contrast, showed a compensatory mechanism, which involved pelvic retraction; presumably as a possible strategy to improve toe clearance of the swing limb. This finding was also consistent with that reported within the literature (Radler et al., 2010). In the present study, all groups walked with a wider step width compared to TD children. Based on the literature, this may have also influenced on the KAM (Stief, Holder, Feja, et al., 2021), and indeed, KAM was the same for the ITF and ITB groups when compared to TD children. Only the ITT group showed a more external rotated hip, a compensatory mechanism to possibly avoid tripping and falling by gaining space between the stance and swinging limb during swing phase. Likewise, only the ITT group had a greater KAM2 than TD children. In the absence of longitudinal cohort studies, the clinical significance of such changes remains unknown. Although some studies have shown that KAM1, rather than KAM2, is associated with KOA and knee pain in adults

(Amin et al., 2004; Miyazaki et al., 2002), there is also evidence that persons with osteoarthritis also present with internal tibial torsion to some degree (Turner, 1994). It is also possible that, as a result of the compensatory external hip rotation during walking, children and adolescents with ITT may be predisposed to patellofemoral pain (Cibulka & Threlkeld-Watkins, 2005). Compensatory mechanisms may exist subconsciously in order to prevent tripping and falling in children and adolescents and are dependent on the location of rotational malalignment (femoral, tibial or both). Moreover, the recognition of the presence of such compensatory mechanisms should aid physicians and researchers in order to optimize treatment planning.

Various idiopathic orthopedic deformities influence KAM. According to study 1, unfavorable deformities which increase KAM1 and therefore medial compartment knee loading were femoral anteversion (Alexander et al., 2019; Bruderer-Hofstetter et al., 2015), external tibial torsion (Alexander et al., 2020) and genu varum (Stief et al., 2011). It was surprising, therefore, that in study 2 of the present thesis, children with femoral anteversion (ITF) showed similar KAM to TD. One potential explanation is that KAM may have normalized in the current study, by children and adolescents with femoral anteversion adopting a greater step width during walking than TD children. Finally, mild idiopathic orthopedic deformities with or without compensatory mechanisms may be present in a healthy population and although they generally do not require treatment (Buldt et al., 2015; Snow, 2021), the findings of the current thesis highlight the importance of considering such factors when conducting research.

The effect of foot posture on KAM has also been greatly discussed within the literature. In general, the literature review (study 1) found that flatfoot deformity decreased KAM during walking (Kothari et al., 2016; Powell et al., 2016). Similarly, in study 3, children and adolescents with flatfoot deformity had lower KAM1 and KAM2 peaks during walking than TD children. In this case, it is conceivable that the load distribution might shift towards the lateral compartment of the knee joint, similar to that reported in persons with a valgus alignment of the knee (Hoch & Weinhandl, 2017), which has been associated with lateral KOA (Brouwer et al., 2007; Felson et al., 2013). It is important to note, however, that there is currently no literature that directly links flatfoot deformity with lateral KOA. Rather, flatfoot deformity has been linked with degenerative change in medial femoral cartilage (Gross et al., 2011) and static measures of "pronated foot posture" was reportedly common in patients with medial compartment KOA (Levinger et al., 2010).

The relationship of the subcomponents of flatfoot deformity to KAM1 and KAM2 were investigated in detail in study 3. The standard parameters of the OFM marker set hindfoot eversion and forefoot abduction were not associated with KAM1 or KAM2 as hypothesized. In contrast, a new parameter calculated from the OFM markers, the lateral calcaneal shift, was found to be moderately correlated with KAM1 and KAM2. In a study included in the systematic review (study 1), foot joint rotations also did not correlate with either peak of KAM (Buldt et al., 2015), with neither hindfoot nor midfoot abduction associated with KAM. This study used a different marker set up to that used in the current thesis, so no direct comparison can be made. However, finding that only lateral calcaneal shift was related to KAM, suggests it might be an important indicator of foot alignment when evaluating KAM rather than more conventional parameters.

An important consideration for studies evaluating KAM, which was most evident in reviewing the literature (study 1) is that there is currently no consensual agreement on how to report KAM and different approaches are used to interpret and display KAM. As early as 1996 (Õunpuu et al., 1996), it was mentioned that there was no existent standard definition of the frontal plane knee moment, setting a base for individual interpretation. KAM can be expressed as either the abduction or adduction moment and as the external or internal moment. The main controversy lies in the inclusion of aspects added to the calculation of the external and internal moment and the direction of numerical values for plotting KAM. For some researchers in the field, moments are already described as internal as soon as body segment acceleration is added; which is included in inverse dynamics calculation (Baker, 2013). External moments, therefore, only consider the relation of the ground reaction force vector to the joint center. For others, however, internal moments must include estimates of muscle force and activity within the calculation (Derrick et al., 2020). The frontal plane knee moment in study 2 and 3 of the present thesis was somewhat arbitrarily defined as the internal knee adduction moment without the inclusion of muscle force estimations.

4.1. Clinical Significance and outlook

Extensive research has examined the impact of various orthopedic deformities of the lower extremity, especially genu varum, on KAM in adults (Brouwer et al., 2007; Sharma et al., 2010), given its potential link to the development of knee pain and degenerative knee joint disease (Amin et al., 2004; Andriacchi & Mündermann, 2006; Baliunas et al., 2002; Mündermann et al., 2005). Considering children, several studies have also been conducted showing the impact of orthopedic deformities on gait biomechanics (Alexander et al., 2019, 2020; Bruderer-Hofstetter et al., 2015; Kothari et al., 2016; MacWilliams et al., 2016; Stief et al., 2011). However, none of these studies have included idiopathic internal tibial torsion, the combined internal femoral and tibial torsion or the subcomponents of idiopathic flatfoot deformity to their analysis. Concerning KAM, it is particularly important to quantify factors relating to KAM in children, in order to follow up with strategies that mitigate against the development of joint disease in later life. Therefore, this thesis aimed to fill specific knowledge gaps of the effects of lower extremity internal rotational malalignment on compensatory mechanisms and their potential effects on KAM and how the subcomponents of flatfoot deformity (hindfoot valgus, forefoot abduction and medial longitudinal arch height) correlate with KAM in children and adolescents during walking.

In the first part of this thesis (Chapter 3.1), a systematic review was undertaken to identify factors that have been reported within the literature to impact upon KAM during clinical three-dimensional gait analysis. The review was broad and important from both a research and clinical perspective, as it not only identified individual, participant-related factors, such as age, weight and idiopathic orthopedic deformities, but also identified modifiable factors that could be influenced by the protocol used to collect gait data, such as walking speed. The latter factors are of critical importance for both clinical and research-related gait protocols to ensure the comparability of findings, particularly for KAM. For instance, the review highlighted the importance of carefully controlling gait speed and normalizing gait data for age-related changes in height and weight, which is of critical importance in prospective studies of children and adolescents where rapid changes in these parameters occur with growth and development. Further, the review aimed to support the interpretation of gait data in clinical settings for therapy and treatment planning, as reasons for changed KAM values can be systematically considered and ruled out. Moreover, the review was also undertaken to aid research planning, and was subsequently used to inform two experimental studies evaluating KAM in children with idiopathic internal rotational malalignment of the lower extremity and flatfoot deformity.

In the second part of this thesis, two experimental retrospective studies were undertaken to address a current knowledge gap concerning 1) the effect of idiopathic internal rotational malalignment of the lower extremity on compensatory gait strategies and their effect on KAM (Chapter 3.2) and 2) the association of the subcomponents of idiopathic flatfoot deformity, such as hindfoot valgus, forefoot abduction, etc., with KAM (Chapter 3.3). It was shown that children and adolescents with internal rotational deformity of the lower limb adopt a number of compensatory mechanisms to counteract in-toeing gait, which tended to normalize KAM1 values compared to those of TD children. Indeed, in-toeing gait was not evident in about one quarter of children with internal rotational deformity of the lower limb; indicating the need for clinicians to include additional assessments of lower limb alignment than relying on gait analysis alone. The observation that compensatory mechanisms tended to normalize KAM1 values to those compared to TD children has important clinical implications, as greater KAM1 has reportedly been linked with degenerative knee joint disease in adults (Andriacchi & Mündermann, 2006; Baliunas et al., 2002; Mündermann et al., 2005; Sharma et al., 1998). Clinicians should be aware, therefore, that compensatory strategies adopted by children with internal rotational malalignment, such as greater step width during walking, may have a potentially protective effect on knee joint loading. Moreover, it should be noted, that only children with internal tibial torsion (ITT) presented with greater KAM2 values compared to TD children. Although the clinical implications of greater KAM2 have not yet been reported, it is possible that, like KAM1, it may influence pain and long-term health of the knee joint. Furthermore, knowledge of the compensatory mechanisms and their potential benefits on KAM is particularly of interest when the primary deformity, e.g. the internal rotational malalignment of the lower limb, is treated. When corrective surgery is indicated and completed, compensatory mechanisms may dissipate once the deformity has been reduced or may still be present due to a kind of automated or "learned" behavior. In any case, non-physiological knee joint loading might be present after correction of the deformity, which should be evaluated and followed in these patients. Overall, it is recommended, that further research, involving a longitudinal study design, is undertaken to evaluate the mid- to long term consequences of greater KAM2, particularly in children and adolescents with internal rotational deformities of the lower limb. In addition, there is also a need for future research to evaluate the impact of corrective treatment on the compensatory mechanisms and on KAM over the long-term, especially during continued growth of the children.

Although the biomechanical effects of idiopathic flatfoot deformity in children has been previously reported to result in a lower KAM2 (Kothari et al., 2016; Twomey & McIntosh, 2012), to date, no research was found that has evaluated the association of both KAM peaks on the individual subcomponents of flatfoot deformity during walking. Therefore, this second experimental study (Chapter 3.3), directly addressed this limitation. As flatfoot deformity is a combination of deformities (Mosca, 2010), it can be subdivided into three main subcomponents: 1) hindfoot valgus, 2) forefoot abduction and 3) low medial longitudinal arch height. This thesis attempted to identify the subcomponent that was most strongly related to KAM. To aid this process, a newly introduced parameter, termed "lateral calcaneal shift" was calculated based on the standard OFM marker set. The measure was designed to reflect the so-called translatory hindfoot valgus (Thompson & Abaza, 2010), as opposed to the more conventional measure of hinge valgus, as it is difficult to distinguish between a translatory and hinge hindfoot valgus in daily clinical work. Importantly, from both a clinical and research perspective, this thesis demonstrated that none of the standard OFM marker set parameters currently used with collection of gait data correlated with KAM in children and adolescents with flatfoot deformity. In comparing the newly developed parameter with conventional measures, however, lateral calcaneal shift was found to be moderately correlated with KAM in this cohort. Moreover, in a sub-group analysis of surgical cases, change in lateral calcaneal shift remained moderately correlated to the change of KAM1 following surgical correction of flatfoot deformity. Hence, it is recommended that lateral calcaneal shift be implemented as a standard measure in research and clinical gait analysis to further elucidate its role in clinical relevance to knee joint pathology. Further research might be helpful to investigate how the potential protective effect on knee joint loading with flatfoot deformity changes into more harmful or pain developing loading when the deformity is treated.

This thesis provides new insights into both the gait strategies used by children to compensate idiopathic internal rotational malalignment of the lower limb and their effect on KAM and how subcomponents of idiopathic flatfoot deformity correlate with KAM. However, further research, including more comprehensive musculoskeletal models, for example, is needed where the parameter KAM could be systematically changed. Considering the effects of the compensatory mechanisms in internal rotational malalignments, future research should be directed towards evaluating whether these mechanisms disappear following derotation osteotomy for correction of the deformity and how a correction may affect a compensatory mechanism which acts to normalize KAM. Furthermore, while this thesis has also provided first evidence of the clinical utility of the newly developed measure, the so-called lateral calcaneal shift, future studies should investigate the parameter in more detail and, in particular its measurement properties by evaluating its accuracy, reliability, and sensitivity in a variety of clinical populations.

5. Conclusion

On the basis of the three studies conducted within this thesis, some suggestions can be made for clinical and research settings that use three-dimensional instrumented gait analysis, and specifically the outcome variable KAM, as part of their routine examinations and decision-making process. In conducting of longitudinal studies involving multiple gait measurements for either therapeutic or observational purposes, changes in individual characteristics (such as age), gait modifications (such as walking speed) and idiopathic orthopedic deformities should be considered when interpretating data in which KAM is evaluated and of relevance. Moreover, as demonstrated in the retrospective studies undertaken in this thesis, compensatory mechanisms might be present in persons with idiopathic orthopedic deformities, as found in children with internal rotational malalignment, which may act to significantly increase or decrease KAM during walking. The present thesis has also shown that, when evaluating foot posture, the newly introduced measure of ‘lateral calcaneal shift’ might be of interest when examining KAM, as an adjuvant to standard foot parameters.

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7. Appendix

7.1. Original Article of Study 1:

Frontal plane knee moment in clinical gait analysis: A systematic review on the effect of kinematic gait changes

S. Kimberly Byrnes[1,2], Jana Holder[3,4], Felix Stief[3,4], Scott Wearing[2], Harald Böhm[1], Chakravarty U. Dussa[1], T. Horstmann[2,5]

[1] Orthopedic Children's Hospital, Kind im Zentrum – Chiemgau, Aschau, Germany

[2] Department of Sports and Health Sciences, Institute for Conservative and Rehabilitative Orthopedics, Technical University of Munich, Germany

[3] Department of Orthopedics (Friedrichsheim), University Hospital Frankfurt, Frankfurt am Main, Germany

[4] Faculty of Medicine, Goethe University, Frankfurt am Main, Germany

[5] Medical Park St. Hubertus Klinik, Bad Wiessee, Germany

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Review

Frontal plane knee moment in clinical gait analysis: A systematic review on the effect of kinematic gait changes

S. Kimberly Byrnes [a,b,*], Jana Holder [c,d], Felix Stief [c,d], Scott Wearing [b], Harald Böhm [a], Chakravarty U. Dussa [a], Thomas Horstmann [b,e]

[a] Orthopedic Children's Hospital, Kind im Zentrum – Chiemgau, Aschau, Germany
[b] Department of Sports and Health Sciences, Institute for Conservative and Rehabilitative Orthopedics, Technical University of Munich, Germany
[c] Department of Orthopedics (Friedrichsheim), University Hospital Frankfurt, Frankfurt am Main, Germany
[d] Faculty of Medicine, Goethe University, Frankfurt am Main, Germany
[e] Medical Park St. Hubertus Klinik, Bad Wiessee, Germany

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ABSTRACT

Introduction: The frontal plane knee moment (KAM1 and KAM2) derived from non-invasive three-dimensional gait analysis is a surrogate measure for knee joint load and of great interest in clinical and research settings. Many aspects can influence this measure either unintentionally or purposely in order to reduce the knee joint load to relieve symptoms and pain. All these aspects must be known when conducting a study or interpreting gait data for clinical decision-making.
Methods: This systematic review was registered with PROSPERO (CRD42020187038). Pubmed and Web of Science were searched for peer-reviewed, original research articles in which unshod three-dimensional gait analysis was undertaken and KAM1 and KAM2 were included as an outcome variable. Two reviewers independently screened articles for inclusion, extracted data and performed a methodological quality assessment using Downs and Black checklist.
Results: In total, 42 studies were included. Based on the independent variable investigated, these studies were divided into three groups: 1) gait modifications, 2) individual characteristics and 3) idiopathic orthopedic deformities. Among others, fast walking speeds (1) were found to increase KAM1; There were no sex-related differences (2) and genu valgum (3) reduces KAM1 and KAM2.
Conclusion: While consistent use of terminology and reporting of KAM is required for meta-analysis, this review indicates that gait modifications (speed, trunk lean, step width), individual characteristics (body weight, age) and idiopathic orthopedic deformities (femoral or tibial torsion, genu valgum/varum) influence KAM magnitudes during walking. These factors should be considered by researchers when designing studies (especially of longitudinal design) or by clinicians when interpreting data for surgical and therapeutic decision-making.

1. Introduction

Deviations in the mechanical load at the knee joint from a normal range may induce musculoskeletal pain [1] and/or stimulate the onset of joint disease and degeneration later in life [2]. The frontal plane moment of the knee during gait is a widely researched and commonly used indicator of mechanical loading of the knee joint [3,4] and often employed in clinical decision-making and therapeutic planning in orthopedics [5–9]. In particular, the peak external knee adduction moment (KAM1) is associated with the rate of progression and initiation of osteoarthritis (OA) [10,11].

The frontal plane knee moment can be derived non-invasively from three-dimensional multi-camera gait analysis systems synchronized with a force platform using an inverse dynamics approach [12]. Most commonly the frontal plane knee moment is expressed as the external knee *adduction* moment (varus moment) [13–17]. Gait compensating strategies have been shown to influence the frontal plane knee moment. For instance, strong associations have been shown between the foot

* Corresponding author at: Gait Laboratory, Orthopedic Hospital for Children, Kind im Zentrum – Chiemgau, Bernauerstr. 18, 83229 Aschau i, Chiemgau, Germany.
E-mail address: k.byrnes@kiz-chiemgau.de (S.K. Byrnes).

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progression angle and KAM. A toe-out gait pattern reportedly reduces the second peak of the KAM waveform, whereas an increased toe-in gait reduces the first peak KAM in healthy individuals [17,18]. Similarly, ipsilateral trunk lean toward the affected stance limb has been identified as an important compensatory mechanism to unload the hip or knee joint and to relieve pain during walking [19,20]. Gait modifications, such as increased step width, hip internal rotation, weight transfer to the medial foot and toe-out gait [21] and footwear [22] have all been specifically noted to influence KAM in healthy adults and osteoarthritic patients and have been previously summarized in systematic reviews.

Clinical decisions especially in pediatric orthopedics are regularly based on the outcome of pathological frontal plane knee moments in combination with clinical examination [5,6,8,9]. Furthermore, different idiopathic deformities, i.e., flatfoot deformity and increased knee valgus, can occur in combination and have an impact on the knee adduction moment [23]. This may influence the therapy decisions. Therefore, the aim of this systematic review is to provide researchers and pediatric clinicians with an overview of factors that may influence the frontal plane knee moment during walking. Since children are normally not subject to osteoarthritis, studies that investigated a healthy asymptomatic population or a population with idiopathic orthopedic deformity such as flatfoot, rotational or angular deformities of the lower limbs were searched. Because gait measurements in pediatric clinical settings are commonly performed barefoot, this overview summarizes knee adduction moment variability during barefoot walking over level ground without any type of footwear, insole, brace or walking aid. Thus, the review focusses on participant-related characteristics rather than technical procedures or laboratory settings. This review may be helpful for the interpretation of the frontal plane knee moment, both when comparing data from one-time measurements of multiple participants as well as comparing data of repeated measurements within one participant.

2. Methods

2.1. Review registration

This systematic review was registered with PROSPERO (# CRD42020187038) and followed the PRISMA guidelines [24].

2.2. Search Strategy

Pubmed and Web of Science databases were searched on June 25th, 2021. The following electronic search strategy was used for both databases with database specific truncations: ("knee adduction moment*" OR KAM OR "knee moment*" OR "knee joint loading" OR "knee loading" OR "knee varus moment*" OR "knee valgus moment*" OR "knee varus torque*" OR "knee valgus torque*" OR "knee torque*") AND ("Gait"[Mesh] OR "Gait Analysis"[Mesh] OR "Walking"[Mesh]).

2.3. Selection criteria

Following duplicate deletion, two reviewers (SKB and JH) screened all titles and abstracts for inclusion. Full texts were inspected if insufficient detail was presented in the abstract. Only peer-reviewed original research articles in English between 1990 and June 2021 were considered for this review. Studies were included if they investigated frontal plane knee moments during barefoot overground walking on a level surface and used three-dimensional kinematic and kinetic measurements. Eligible studies presented follow-up or only one-time measurements. Studies were limited to those that reported healthy, asymptomatic persons (i.e. reported no medical history of neurological or musculoskeletal disorders, such as degenerative joint diseases). No restrictions were placed on participant age or sex and studies reporting populations with idiopathic orthopedic deformities, such as flatfoot, leg axis or rotational lower limb deformities were also included. Studies that evaluated the influence of technical (e.g. shoe modifications, insoles, prostheses) or surgical procedures (e.g. osteotomies around the knee due to axial deformities) on the knee joint moment were not included. Conference proceedings, meeting abstracts and dissertations were also excluded from this review.

Disagreements in study selection were discussed and resolved between reviewers; a third reviewer was not involved. Authors of studies were contacted if important information concerning inclusion was not reported.

2.4. Data extraction and quality assessment

A customized form was used for data extraction. One reviewer (SKB) extracted the data. Quality assessment was based on a checklist introduced by Downs and Black [25] with the modifications adopted from Schmid and colleagues [26]. For this systematic review, however, a clear description of the frontal knee moment (internal vs. external and abduction vs. adduction moment) was necessary to generate an overview and aid in the comparison of the results. Hence, an additional item was added to the *reporting* section (2a: Was the parameter "frontal knee moment" clearly described: yes = 1, no = 0). Similarly, Item 4 ("description of intervention was clearly described"), which was removed in the adapted checklist by Schmid et al. [26], was retained for quality assessment as outlined in the original checklist by Downs and Black [25] (Table 1). Different gait modification strategies, for example, were regarded as an intervention. These were assessed with Item 4 as to how the interventions were described and executed in the included studies. Thus, the maximum score in this quality assessment category was 12 points distributed over 10 items. As outlined by Schmid et al. [26], other assessment categories including external validity, internal validity, confounding factors and power were retained. Two reviewers (SKB, JH) independently assessed all the eligible records for their quality.

2.5. Parameter frontal plane knee moment

The parameter describing the frontal plane knee moment was extracted exactly as reported in the records. However, given the wide variety in terminology and in order to minimize confusion, this review adopted the definitions of the International Society of Biomechanics [14] in which all positive reported numerical values derived from inverse dynamics are defined as (external) knee *adduction* or *varus* moments, and negative values as (external) knee *abduction* or *valgus* moments (Fig. 1). In several instances, internal moments were reported with negative values, and subsequently were converted to external moments by assuming a stable joint system in which external and internal moments were balanced [14]. Hence, the term KAM (knee adduction moment) has been used throughout the text.

2.6. Statistical analysis

All analyses were performed using the Statistical Package for Social Science (Version 26, IBM Coporation, Amonk, NY, USA). Agreement in the quality assessment between raters was evaluated using Cohen's kappa statistic. According to Landis and Koch [27], a kappa value of 0–0.2 describes agreement as "slight", 0.21–0.4 as "fair", 0.41–0.6 as "moderate", 0.61–0.8 as "substantial" and 0.81–1.0 as "almost perfect".

3. Results

3.1. Study selection

The search strategy initially identified 2090 studies in total, with 1425 retained after removal of duplicates (Fig. 2). Of those retained, 285 were found to meet the inclusion criteria based on initial title screening. After abstract screening, 108 full-text articles were assessed. In 15 of

Table 1
Quality assessment based on Downs and Black [25] and partly adopted by Schmid et al. [26] (n = 42).

		Reporting (n = 13)		External validity (n = 3)		Internal validity (n = 3)		Internal validity; confounding (n = 2)		Power (n = 2)		Total score (n = 23)	
Author	Year	*SKB*	*JH*	*SKB*	*JH*	*SKB*	*JH*	*SKB*	*JH*	*SKB*	*JH*	*SKB*	*JH*
Alexander et al.	2020	12	12	2	1	3	3	1	1	1	2	19	19
Alexander et al.	2019	13	12	2	2	3	3	1	1	0	0	19	18
Anderson et al.	2018	13	13	2	2	2	3	1	1	0	0	18	19
Ardestani	2016	12	12	2	2	3	3	2	2	0	0	19	19
Bruderer et al.	2015	12	12	2	1	3	3	1	1	0	0	18	17
Buldt et al.	2015	11	11	2	2	3	3	1	1	0	0	17	17
Byrnes et al.	2020a	11	12	2	2	3	3	1	1	0	0	17	18
Byrnes et al.	2020b	12	12	2	2	3	3	1	1	0	0	18	18
Cho et al	2004	12	11	1	1	3	3	1	1	0	0	17	16
Cui et al.	2019	9	9	1	1	3	3	1	1	0	0	14	14
Davids et al.	2014	10	10	2	2	2	3	1	1	0	0	15	16
Farr et al.	2016	9	9	2	1	3	3	1	1	0	0	15	14
Farr et al.	2014	10	10	2	2	3	3	1	1	0	0	16	16
Ferrigno et al.	2016	12	12	2	2	3	3	1	1	1	1	19	19
Fischer et al.	2016	11	11	1	1	3	3	1	1	0	0	16	16
Kerrigan et al.	2000	10	9	2	2	3	3	1	1	0	0	16	15
Kothari et al.	2016	8	8	2	2	3	3	1	1	0	0	14	14
Kulmala et al.	2013	13	11	1	1	3	3	1	1	0	0	18	16
Longpré et al.	2013	12	12	2	2	3	3	2	1	0	0	19	18
Lynn et al.	2008	10	11	2	2	3	3	1	1	0	0	16	17
MacWilliams et al.	2010	11	10	2	2	3	3	1	1	0	0	17	16
MacWilliams et al.	2016	10	11	2	2	2	3	1	1	0	0	15	17
Mahaffey et al.	2018	10	10	2	2	3	3	0	0	0	0	15	15
McMillan et al.	2010	9	9	2	1	3	3	1	1	0	0	15	14
McMillan et al.	2009	11	10	2	2	3	3	1	1	0	0	17	16
Nagano et al.	2020	9	10	2	2	3	3	0	0	0	0	14	15
Ota et al.	2015	12	11	2	2	1	3	1	1	0	0	16	17
Pamukoff et al.	2016	12	11	2	2	3	3	1	1	1	2	19	19
Powell et al.	2016	13	13	3	2	3	3	1	1	0	0	20	19
Robbins et al.	2009	12	12	2	3	3	3	0	0	0	0	17	18
Robbins et al.	2016	12	11	2	2	3	3	2	1	1	2	20	19
Schwartz et al.	2008	8	8	1	1	3	2	2	1	0	0	14	12
Shultz et al.	2009	11	11	2	2	3	3	1	1	0	0	17	17
Stevens et al.	2004	10	9	1	1	3	3	1	1	0	0	15	14
Stief et al.	2011	13	13	1	1	3	3	1	1	0	0	18	18
Stief et al.	2021	13	13	1	1	3	3	1	1	0	0	18	18
Street & Gage	2013	13	13	1	1	3	3	1	1	0	0	18	18
Sun et al.	2018	13	13	1	1	2	3	1	1	0	0	17	18
Teichtahl et al.	2006	12	12	2	2	3	3	1	1	0	0	18	18
Teichtahl et al.	2009	12	12	2	2	3	3	1	1	0	0	18	18
van den Noort et al.	2013	13	12	2	2	3	3	1	1	0	0	19	18
van der Linden	2002	11	10	1	1	2	3	1	1	0	0	15	15

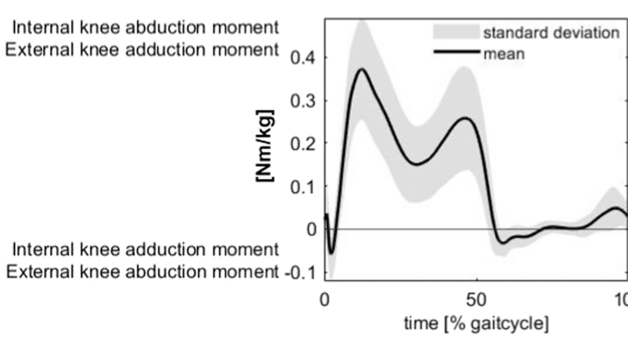

Fig. 1. A general illustration of pseudo data to clarify the definition of knee adduction moment in this systematic review. The external knee *adduction* moment is shown with positive values.

these studies, it was not clearly described if participants were analyzed during barefoot or shod gait. Authors of these studies were contacted via email, and eleven authors replied within the given period of eight weeks. Six of these studies evaluated shod gait and were subsequently excluded. Four studies were excluded as no response was received from the authors. After full-text screening a total of 34 eligible studies were included (Fig. 2). Additional review of the reference list from three systematic reviews yielded further three articles for inclusion. Similarly, after screening the reference list of the 37 eligible studies, five additional studies were included. Hence, a total of 42 studies were included in this systematic review.

3.2. *Quality assessment*

Cohen's kappa statistic revealed an almost perfect agreement in the quality assessment (Table 1) between reviewers ($\kappa = 0.894$, $p < .001$). No study received the total score of 23. The highest score was 20 for two studies [16,28]. One study achieved a score of only 12 [29]. The average score of all studies was 16.

3.3. *Methodology and terminology*

Adults and children participated in the included studies (Table 2). In total, 22 studies analyzed adults, 17 of these included men and women [15–17,28,30–42], 3 only men [43–45] and 2 only women [46,47]. The other 20 studies analyzed children, 18 of these included boys and girls [5–9,13,23,29,48–57] and 2 only boys [58,59].

Frontal plane knee moment was reported as the external KAM in 19 studies [13,15–17,31,32,35,38,40–43,46–49,55–57], the internal KAM in two studies [50,51] and six described the internal knee abduction moment [28–30,39,44,54]. Four studies used the term "internal valgus

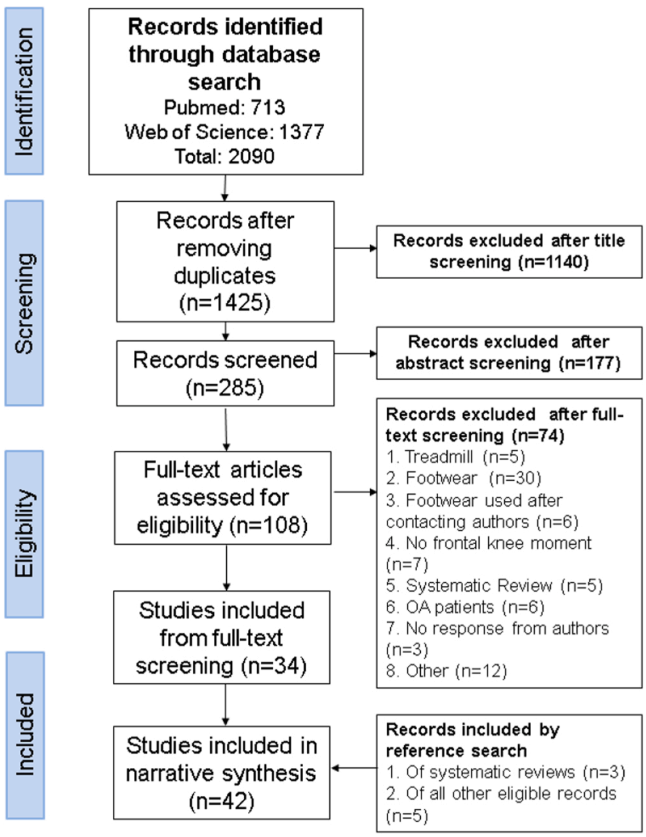

Fig. 2. Flow Chart of study inclusion based on Moher et al. [24].

knee moment" [5,6,8,52], one used "internal knee varus moment" [23] and in nine studies the frontal plane knee moment was not clearly defined [7,9,33,34,36,45,53,58,59]. Both peaks of the frontal plane knee moment were investigated in 25 studies [5,15–17,23,28–30,32–36,38,41,42,44,46,47,50–52,55–57]. Seven studies reported the maximum frontal plane knee moment that occurred during the first half of stance phase [6,37,39,40,43,45,54]. Three studies reported the knee moments at gait events defined by the opposite limb [7–9], two studies presented the peak amplitude during first and second half of stance phase [53,58] and five studies used principle component analysis or statistical parametric mapping to analyze the complete gait cycle [13,31,48,49,59].

The majority of studies normalized the frontal plane knee moment to body weight, reporting values in the units of Nm/kg [5–9,13,15,16,28–30,41,43,44,46,48–52,55]. Ten studies normalized the knee moment to body weight and body height, reporting frontal plane knee moments as percentage of the product of body weight and body height (%bw*bh) [17,31,32,34,35,39,42,45,47,56]. Less commonly frontal plane knee moments were reported in units of Nm/kgm [33,36,38,53,57,58] or Nmm/kg [23]. Three studies did not normalize the frontal plane knee moment to body weight and body height, hence, the unit reported was Nm [40,54,59].

3.4. Influences on the frontal plane knee moment

Depending on the independent variables of interest within each study, studies were broadly divided into three overall groups: (1) gait modifications, (2) individual characteristics, and (3) idiopathic orthopedic deformities (Fig. 3).

3.4.1. Gait modifications

Gait modifications were defined as intentional actions taken by a participant during gait analysis measurement. Among these were studies that investigated differences in gait speed [17,29,31,40,43,54,57], the effect of lateral trunk lean [16,17,30], changes in step width [41,42], changes in foot progression angle (toeing-in or toeing-out) [15,17,34,42], a change to a medial thrust knee gait pattern [35], a draw-in maneuver[38] or knee extensor or flexor dominant gait patterns [44] (Table 2).

Most studies evaluating gait speed reported that walking at speeds slower than preferred or freely selected speed had no significant effect on frontal plane knee moment [17,29,40,43]. Faster than preferred gait speeds, in contrast, typically increased KAM1 [17,29,31,43]. KAM2 was found to be increased in two studies [17,57], whereas one study found that very fast speeds reduced KAM2 compared to slow walking [29]. However, one study found that the influence of gait speed on KAM depended on how the faster gait speed was attained. Faster walking speeds arising from an increase in cadence only did not result in a change in KAM1 [31], while those achieved by increasing stride length significantly increased the first peak in KAM [31]. Walking at speeds that were faster than preferred speeds and achieved by an increase in both cadence and stride length also significantly increased KAM1 [31].

Intentional lateral trunk lean towards the ipsilateral side during gait showed significant reductions to KAM in the ipsilateral limb, especially the first peak [16,17,30]. Anderson and colleagues[30] reported that some people increased their step width in order to achieve greater lateral trunk lean, which may also result in reduced KAM. Indeed, Stief et al. [41] reported that a wider than habitual step width reduced KAM1 and KAM2.

In-toeing was associated with a reduction in KAM1 in two studies [17,34] and an increase in KAM2 in one study [15]. Out-toeing, in contrast, had the opposite effect, reportedly increasing KAM1 in one study [17] but reducing KAM2 in two studies [15,17] and more notably in the non-dominant limb [42]. Cui et al. [34], however found no differences in KAM with toe-out compared to natural walking, when participants were asked to achieve a 90° angle between the crossing toe-heel lines of left and right foot.

A medial knee thrust gait reduced KAM2 [35] while a decrease in thoracic kyphosis which can be achieved by drawing in the core/belly while walking reduced KAM1 [38]. KAM1, however, was increased for men with a knee extensor dominant gait pattern and KAM2 was increased for men with a knee flexor dominant gait pattern [44] (Table 3).

3.4.2. Individual characteristics

Included studies in this group investigated individual characteristics, such as sex [33,36], age [37], limb dominance[42,56], body weight [39,45,53,54,58,59] and neuromuscular fatigue [46] (Table 2). Sex and neuromuscular fatigue did not alter KAM significantly [33,36,46]. Greater age, on the other hand increased KAM1 when elderly versus young adults were compared [37]. Regarding limb dominance, which was determined by the limb that was first used to step when gait was initiated, one study showed significant increases in KAM of the dominant compared to the non-dominant limb [42], whereas the other study found no difference in KAM between the dominant and non-dominant limbs [56]. Concerning body weight, one study investigated body weight unloading and found significant reductions in KAM1 with bodyweight unloading (of 15 % and 30 %) compared to normal body weight [45]. The remaining five studies compared overweight and healthy weight cohorts. One study found no significant difference in KAM between overweight and healthy weight participants [39]. Shultz and colleagues [54] found significantly greater absolute peak KAM in overweight adults, however, this difference was not significant when KAM was normalized to body weight. Moreover, high body fat was positively associated with greater KAM during the middle of stance [59]. Finally, two studies by McMillan et al. reported a lower peak KAM amplitude in obese children compared to healthy weight children [53,58] (Table 3).

Table 2
Summary of independent variable, outcome measures and results of reviewed studies.

Group	Author	Parameter	Maximum (max) or first and second peak (1&2)	Adduction (AD) or Abduction (AB) Internal (int) or external (ext) moment	Unit	Results
Gait modifications	Ardestani 2016	gait speed	SPM	AD ext	% bw*bh	KAM1 sig. greater when stride length was increased for FW KAM1 sig. greater when both (stride length & cadence were increased) for FW
	Robbins 2009	gait speed	max	AD ext	Nm	KAM1 sig. greater in FW compared to SW
	Schwartz 2008	gait speed	1&2	AB int	Nm/kg	KAM1 sig. greater in FW compared to SW KAM2 sig. reduced in FW compared to SW
	Shultz 2009	gait speed	max	AB int	Nm	no sig differences between speeds
	Sun 2018	gait speed	max	AD ext	Nm/kg	KAM1 sig. greater in FW compared to NW
	van den Noort 2013	gait speed	1&2	AD ext	% bw*bh	KAM1 sig. greater in FW compared to NW KAM2 sig. greater in FW compared to NW
	van der Linden 2002	gait speed	max	AD ext	Nm/ kgm	KAM1 sig. greater in FW compared to NW KAM2 sig. greater in FW compared to NW
	Anderson 2018	lateral trunk lean with or without changed step width	1&2	AB int	Nm/kg	KAM1 sig. reduced with LTL with WSW strategy KAM2 sig. reduced with LTL with WSW strategy KAM1 sig. reduced with LTL with NSW strategy
	Robbins 2016	lateral trunk lean	1&2	AD ext	Nm/kg	KAM1 sig. reduced with LTL (decreased gait speed as well during LTL gait)
	van den Noort 2013	lateral trunk lean	1&2	AD ext	% bw*bh	KAM1 sig. reduce in LTL compared to NTL
	Street 2013	narrow step width and limb dominance	1&2	AD ext	% bw*bh	KAM1 sig. greater in dominant limb with narrow step compared to non-dominant limb KAM2 sig. greater in dominant limb with narrow step width compared to non-dominant limb
	Cui 2019	toe-in	1&2	AD	% bw*bh	KAM1 sig. reduced with toe in gait
	Lynn 2008	toe-in	1&2	AD ext	Nm/kg	KAM2 sig. greater with toe in gait
	van den Noort 2013	toe-in	1&2	AD ext	% bw*bh	KAM1 sig. reduced with toe in gait compared to normal
	Cui 2019	toe-out	1&2	AD	% bw*bh	no sig. differences between toe out gait and normal gait
	Lynn 2008	toe-out	1&2	AD ext	Nm/kg	KAM2 sig. reduced with toe out gait
	Street 2013	toe-out	1&2	AD ext	% bw*bh	KAM2 sig. reduced with toe out gait in non-dominant limb compared to dominant limb
	van den Noort 2013	toe-out	1&2	AD ext	% bw*bh	KAM1 sig. greater with toe out gait compared to normal KAM2 sig. reduced with toe out gait compared to normal
	Stief 2021	step width	1&2	AD ext	Nm/kg	KAM1 sig. reduced with WSW compared to habitual step width KAM2 sig. reduced with WSW compared to habitual step width
	Ferrigno 2016	med. knee thrust	1&2	AD ext	% bw*bh	KAM2 sig. reduced with medial thrust gait compared to normal gait
	Ota 2015	draw-in maneuver	1&2	AD ext	Nm/ kgm	KAM1 sig. reduced in thoracic kyphosis (draw-in maneuver) compared to normal gait
	Kumala 2013	knee extensor dominant gait pattern	1&2	AB int	Nm/kg	KAM1 sig. greater in people with knee extensor dominant gait pattern compared to typical gait pattern
	Kumala 2013	knee flexor dominant gait pattern	1&2	AB int	Nm/kg	KAM2 sig. greater in people with knee flexor dominant gait pattern compared to typical gait pattern
Individual characteristics	Cho 2004	sex differences	1&2	AB	Nm/ kgm	no sig. differences
	Kerrigan 2000	sex differences	1&2	AD	Nm/ kgm	no sig. differences
	Street 2013	limb dominance	1&2	AD ext	% bw*bh	KAM1 sig. greater in DL compared to NDL KAM2 sig. greater in DL compared to NDL
	Teichtahl 2009	limb dominance	1&2	AD ext	% bw*bh	no sig. differences
	Fischer 2016	body weight	max	AD	% bw*bh	KAM1 sig. greater in 0% compared to 15&30% body weight unloading KAM1 sig. greater in 15% compared to 30% body weight unloading
	Mahaffey 2018	body weight	PCA	AD ext	Nm	KAM mid stance greater with higher body fat
	McMillan 2009	body weight	1&2 amplitude	AB	Nm/ kgm	KAM1 and KAM2 sig. lower in OW compared to HW
	McMillan 2010	body weight	1&2 amplitude	AB	Nm/ kgm	KAM1 and KAM2 sig. lower in OW compared to HW
	Pamukoff 2016	body weight	max	AB int	bw*bh	no sig. differences between OW and HW

(continued on next page)

Table 2 (*continued*)

Group	Author	Parameter	Maximum (max) or first and second peak (1&2)	Adduction (AD) or Abduction (AB) Internal (int) or external (ext) moment	Unit	Results
	Shultz 2009	body weight	max	AB int	Nm	KAM1 sig. greater in OW compared to HW no sig. differences when normalized to bodyweight
	Nagano 2020	age	max	AD	Nm	KAM1 sig. greater in older compared to younger people
	Longpre 2013	neuromuscular fatigue	1&2	AD ext	Nm/kg	no sig. differences
Idiopathic orthopedic deformity	Alexander 2019	femoral anteversion	PCA	AD ext	Nm/kg	KAM1 sig. greater for femoral anteversion patients KAM2 sig. lower for femoral anteversion patients compared to TD
	Bruderer-Hofstetter 2015	femoral anteversion	PCA	AD ext	Nm/kg	subgroup neutral alignment: KAM sig. greater in mid to terminal stance subgroup valgus alignment: KAM sig. lower during pre-swing
	MacWilliams2016	femoral anteversion	1&2 at opposite gait events	AB	Nm/kg	KAM1 sig. lower for femoral anteversion patients comapred to TD
	Davids 2014	internal tibial torsion	1&2 at opposite gait events	AD	Nm/kg	KAM1 sig. reduced with tibial torsion compared to TD KAM2 tends to be greater with tibial torsion compared to TD
	MacWilliams 2010	internal tibial torsion	1&2 at opposite gait events	valgus knee moment int	Nm/kg	KAM2 sig. greater with internal tibial torsion compared to TD
	Byrnes 2020a	femoral and tibial internal torsion	1&2	AD int	Nm/kg	KAM2 sig. greater with tibial internal torsion compared to TD
	Alexander 2020	external tibial torsion	PCA	AD ext	Nm/kg	KAM1 greater compared to TD KAM2 lower compared to TD
	MacWilliams 2010	external tibial torsion	1&2 at opposite gait events	valgus knee moment int	Nm/kg	KAM2 sig. lower with tibial external torsion compared to TD
	Teichtahl 2006	foot and thigh rotation	1&2	AD ext	% bw*bh	Degree of foot rotation correlated sig. with magnitude KAM2 "women who walk with ext foot rot reduce their KAM2" no sig. correlations between KAM1 and foot rot. or KAM1/KAM2 and thigh rot.
	Farr 2017	genu valgum	1&2	AB int	Nm/kg	KAM2 sig. lower in genu valgum patients compared to TD
	Farr 2014	genu valgum	1&2	AB int	Nm/kg	KAM1 and KAM2 sig. lower in genu valgum patients compared to TD
	Stevens 2004	genu valgum	max	AB int	Nm/kg	KAM1 sig. lower in genu valgum patients compared to TD
	Stief 2011	genu varum	1&2	AD ext	Nm/kg	KAM1 sig. greater in genu varum patients compared to TD KAM2 sig. greater in genu varum patients compared to TD
	Buldt 2015	low, high and neutral arch	1&2	AD ext	% bw*bh	no sig. differences
	Byrnes 2020b	foot posture	1&2	AD int	Nm/kg	KAM1 sig. lower with flat foot compared to TD KAM2 sig. lower with flat foot compared to TD
	Kothari 2016	low and neutral arch	1&2	AD int	Nmm/kg	KAM2 sig. lower in low arch compared to TD
	Powell 2016	low and high arch	1&2	AB int	Nm/kg	KAM1 sig. lower in high arch compared to low arch KAM2 sig. lower in high arch compared to low arch

KAM: knee adduction moment; PCA: principal component analysis; SPM: statistical parametric mapping; bw: body weight; bh: body height; FW: fast walking; SW: slow walking; NW: normal walking; LTL: lateral trunk lean; NTL: normal trunk lean; WSW: wide step width; NSW: normal step width; DL: dominant limb; NDL: non-dominant limb; OW: overweight; HW: healthy weight; TD: typically developed children.

3.4.3. Idiopathic orthopedic deformities

Studies in this group investigated one or more of the following idiopathic orthopedic deformities: increased femoral anteversion [9,48–50], internal tibial torsion [7,8,50], external tibial torsion [8,13], foot and thigh torsion [47], genu valgum [5,6,52], genu varum [55] and foot posture [23,28,32,51] (Table 2). The results of studies investigating increased femoral anteversion were mixed; with one study showing KAM1 was lower for children with increased femoral anteversion compared to children without [9], another showed a higher KAM1 and a lower KAM2 in children with increased femoral anteversion [48] and a third study reporting no difference in KAM1 or KAM2 [50]. Children with neutral alignment and increased femoral anteversion had a higher KAM from mid to terminal stance phase whereas children with additional knee valgus alignment had a decreased KAM during pre-swing phase compared to typically developing children [49].

Increased internal tibial torsion significantly reduced KAM1 [7] and increased KAM2 [8,50] compared to typically developed children. Increased external tibial torsion on the other hand had the opposite effect and reduced KAM2 [8]. In another study, some children with increased external tibial torsion showed compensatory hip internal rotation [13]. Here, KAM was changed dependent on whether children showed compensatory hip rotation or not. Children without compensatory hip rotation had lower KAM1 and KAM2 than those with compensatory hip rotation, who in turn had a greater KAM1 compared to healthy children. Moreover, the degree of clinical external foot rotation correlated with the reduction in KAM2, whereas clinical thigh rotation could not be associated with a change in KAM [47].

Knee valgus alignment reduced KAM1 [6,52] and KAM2 [5,52]

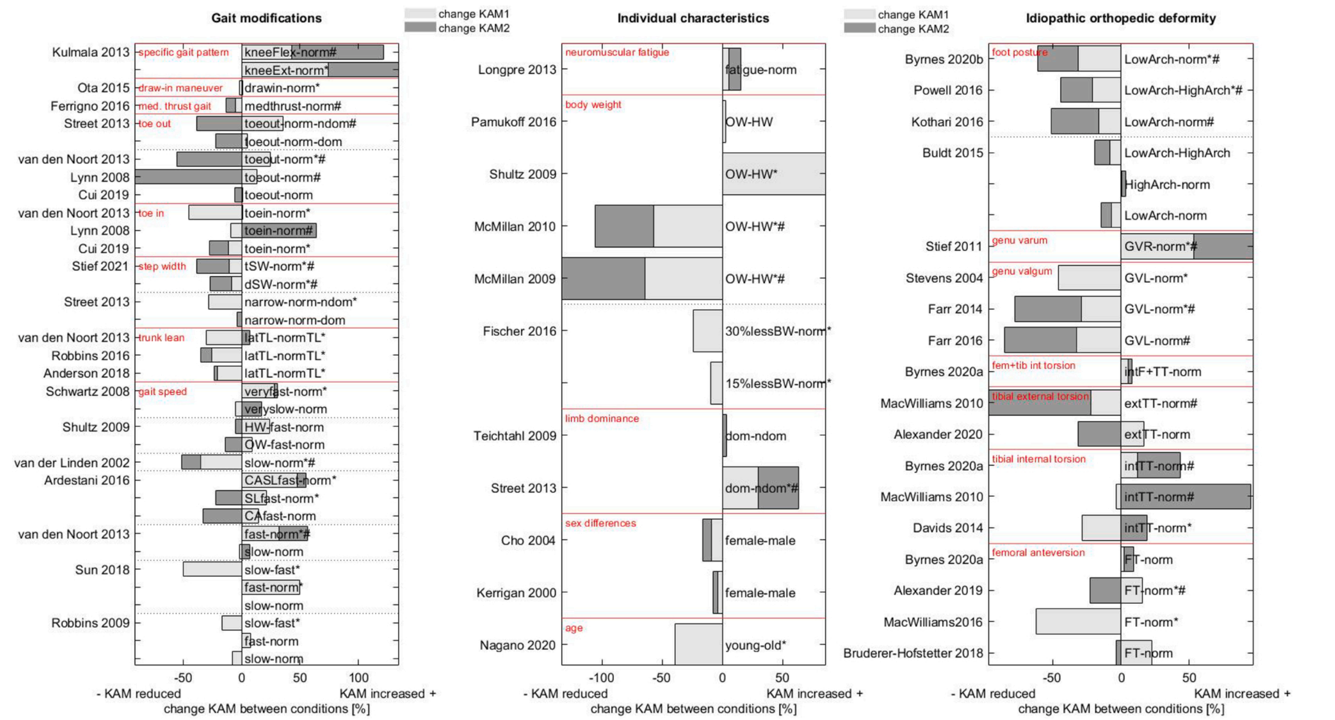

Fig. 3. Summary of the findings of the included studies. Given that some studies did not report absolute values, values were estimated and extracted from graphs where possible. Two studies were not included due to missing graphs and values [56,59]. Shown are the results as the change of the knee adduction moment in per cent between conditions, e.g., toe-in to norm values. The length of the bar is the sum of KAM1 and KAM2. Significant difference of first peak reported in the records were marked with * and second peak with #. Abbreviations: kneeFlex: knee flexor dominant gait pattern; kneeExt: knee extensor dominant gait pattern; tSW: tripled step width; dSW: doubled step width; ndom: non-dominant limb; dom: dominant limb; latTL: lateral trunk lean; normTL: normal trunk lean; HW: healthy weight; OW: overweight; CASLfast: fast walking by increased cadence and stride length; SLfast: fast walking by increased stride length; CAfast: fast walking by increased cadence; BW: bodyweight; GVR: genu varum; GVL: genu valgum; extTT: tibial external torsion; intTT: tibial internal torsion; FT: femoral torsion (anteversion); intF+TT: internal femoral and tibial torsion.

Table 3
Summary of the effects of gait modifications, individual characteristics and orthopedic deformities on first and second peak knee adduction moments.

	First peak knee adduction moment (KAM1)		Second peak knee adduction moment (KAM2)	
	↑ increase	↓ decrease	↑ increase	↓ decrease
Gait modification	Fast walking [17,29,31,40,43,57] Dominant limb with narrow step width [42] Toe-out [17] Knee extensor dominant gait pattern [44]	Lateral trunk lean [16, 17,30] Toe-in gait [17,34] Step width [41] Draw-in strategy [38]	Toe-in gait [15] Knee flexor dominant gait pattern [44]	Fast walking [29] Lateral trunk lean with wide step width [30] Toe-out gait [15] Step width [41] Medial knee thrust [35]
Individual characteristics	Dominant limb [42] Elderly persons compared to younger adults [37]	Body weight unloading [45] Overweight [53,58]	Dominant limb [42]	Overweight [53,58]
Idiopathic orthopedic deformities	Femoral anteversion [48] External tibial torsion [13] Genu varum [55]	Femoral anteversion [9] Internal tibial torsion [7] Genu valgum [6,52] Low arch [51] High arch [28]	Internal tibial torsion [7,8,50] Genu varum [55]	Femoral anteversion [48] External tibial torsion [8,13] Genu valgum [5,52] Low arch [23,51] High arch [28]

compared to a neutral alignment. In contrast, children with genu varum, showed an increased KAM1 and KAM2 [55].

Finally, foot posture, categorized as low, high and neutral-arch, showed significant effects in three out of four studies. No differences in KAM were found in one study between adults with low, high and neutral foot posture [32]. Other studies found that KAM1 [51] and KAM2 [23,51] was reduced in children with a low arch. On the other hand, KAM1 and KAM2 were found to be greater in women with a lower arch compared to women with a higher arch foot posture [28] (Table 3).

4. Discussion

This systematic review summarizes the factors influencing the frontal plane knee moment during barefoot walking. The 42 studies were grouped into three categories: gait modifications, individual characteristics and idiopathic orthopedic deformities according to the independent variable investigated and its effect on the maximum or the first and second peak frontal plane knee moment, referred to as KAM1 and KAM2. As the frontal plane knee moment can be used as a surrogate measure of knee joint loading [3,4] and has been associated with knee osteoarthritis [2,11], the parameter is of great interest in the field of clinical biomechanics. Hence, studies were selected based on aspects that can be present during clinical gait analysis measurement.

4.1. Frontal plane knee moment – the parameter

It was already mentioned in 1996, that researchers use different approaches for interpreting and plotting the frontal plane knee moment [60]. To this day there is no consensus on how to report the frontal plane knee moment. This inconsistency in presenting the frontal plane knee moment made precise and direct comparison or meta-analysis between studies impossible. The main controversy lies in the definition of an internal and external moment and the direction of numerical values (negative or positive direction). While for some researchers an internal moment implies the involvement of muscle force and activity to counterbalance the external forces [14], for others the internal moment already takes the addition of body segment acceleration into account which is included within the inverse dynamics calculation [61]. Whereas for the external moments only the relation of ground reaction force to the joint center is considered. Since the majority of the included studies used the Vicon motion system (n = 28) and the Plug-in Gait model (n = 15), the direction of numerical values was defined based on the Plug-in Gait reference guide where positive values describe knee adduction or external varus moments [12]. Considering that external and internal moments must be equal for a balanced and stable joint system [14], external KAM (positive direction) were set equal to internal knee abduction moments (positive direction) in this review.

4.2. Gait modifications

Intentional gait modifications have been clinically recommended as a strategy to reduce KAM [21] in patients with knee pain and/or osteoarthritis with a view to possibly delaying surgery. Gait speed is a widely known influential parameter. Faster than normal gait speeds had more influence on the knee moment, especially increasing KAM1, while slower than normal gait speeds had less influence on KAM. Faster speeds can be achieved by increasing cadence and/or stride length. Interestingly, the mode of how fast speeds were achieved impacted KAM. Fast walking due only to an increase in cadence had no impact, whereas increased stride length whether in isolation or combined with an increase in cadence combined, significantly increased KAM [31]. Particularly, for comparison of KAM between and within participants a similar walking speed should be achieved for all measurements. However, it may not be necessary to control exactly by a metronome, since this could evoke deviations from habitual gait patterns [62]. A discrepancy of around 5 % or 10 % or 0.1 m/s between gait speeds may be considered reasonable and has already been used in common practice [30,32,63]. Furthermore, if gait changes, such as in- and out-toeing or lateral trunk lean are observed during gait analysis, then their influence on KAM should be considered. Trunk lean was found to reduce KAM, especially KAM1 [16,17,30]. As for the strategy to change the foot progression angle to lower KAMs, both in- and out-toeing can be effectively used as previously described in a different systematic review [18]. Summarizing from studies included in the current review, KAM1 can be reduced with toe-in gait [17,34], whereas KAM2 may be reduced by out-toeing gait [15,17]. Further, a more external foot progression angle (out-toeing) might be adopted with a wider step width gait pattern, to achieve a reduced knee moment [41].

Subtle gait modifications may go un-noticed by practitioners but still influence the outcome on the frontal plane knee moment. For instance, drawing the belly in slightly (reducing anterior pelvic tilt) while walking in order to decrease thoracic kyphosis may not be visible to the examiner but may still affect KAM [38]. Furthermore, as gait can be like an individual finger print [64], person-specific gait patterns have shown changes in KAM. For example, knee extensor dominant gait pattern in male participants increased KAM1 and knee flexor dominant gait pattern increased KAM2 [44]. These gait patterns were defined after persons with an appearance of a knee extensor moment during the entire stance phase or showed dominance of the knee flexor moment during terminal stance phase, respectively [44]. Underlying reasons for the appearance of gait modifications may also be related to an idiopathic deformity, for example when in- or out-toeing occur due to increased or decreased femoral and/or tibial torsion. These are further discussed below in the section of idiopathic deformities. It is also possible that gait modifications might occur intentionally, as an automated gait pattern of the individual, or may be compensatory to avoid pain during walking. Thus, kinematics of gait and the individual's condition (musculoskeletal build, pain, existing injuries etc.) should be considered when evaluating and interpreting or comparing KAM.

4.3. Individual characteristics

Regarding individual characteristics, a decrease in KAM was observed in overweight children compared to healthy weight children and could be explained by the frequent presence of an increased knee valgus angle in overweight children [53,58]. One study found significantly greater KAM in overweight children, however, these differences were removed after normalizing KAM to body weight [54]. Furthermore, body weight unloading (15% and 30% of body weight) affected the knee moment in reducing KAM compared to normal weight [45]. Consideration of changes in body weight may be of interest especially during longitudinal studies where potential weight gain or loss could occur. Regarding limb dominance, two records were included in this systematic review with conflicting outcomes. One study showed an effect on both peaks of KAM [42], where KAM1 and KAM2 were greater in the dominant limb compared to the non-dominant limb and the other found no effect [56]. Higher age was associated with greater KAM1 [37], which may be pertinent to the development of osteoarthritis in the elderly population, where the prevalence for OA is the greatest [65].

Interestingly, this review found that neuromuscular fatigue had no influence on KAM [46]. This might imply that there is no need for patients to abstain from vigorous physical activity immediately prior to gait analysis as required by many laboratories, since it may not influence the outcome of the frontal plane knee moments. It should be noted however, that fatigue did influence sagittal plane knee kinetics [46]. Finally, two studies agreed that sex did not impact KAM [33,36]. Although one of them found significant differences in kinematics, such as narrower step width, a greater knee valgus angle and a more flexed, adducted and internally rotated hip in females [33]. All these factors were shown to possibly influence KAM [5,9,41,48,49,52].

4.4. Idiopathic orthopedic deformities

Different idiopathic orthopedic deformities, either rotational, angular or foot deformities were found to influence the KAM. Considering that a greater KAM1 is associated with joint disease and development of medial knee OA [10,11], the following idiopathic orthopedic deformities may not be favorable, as these increase KAM1: femoral anteversion [48,49], external tibial torsion [13] and genu varum [55]. Compensatory mechanisms should be considered when evaluating orthopedic deformities, as they may occur in order to compensate for the deformity [13,50]. In these cases, therapeutic and surgical planning should be carefully assessed as the outcomes may differ depending on whether compensatory mechanisms remain after the intervention.

Overall, mild idiopathic orthopedic deformities with or without compensatory mechanisms may be present in a healthy population and generally do not require treatment [32,66]. Therefore, the influence of these deformities should be considered especially when evaluating and interpreting the data of healthy participants. Additionally, as mentioned above, in- and out-toeing gait showed influence on KAM [18]. Likewise, femoral and tibial torsion, which may lead to increased or decreased foot progression angle (i.e., in-/out-toeing gait), also had an effect on the direction of KAM.

On a side note, different statistical methods used could potentially lead to different outcomes between studies. However, different results were also apparent with the same statistical method used. In this systematic review statistical methods were not looked at closely and were therefore not included in the discussion of the results.

5. Limitations

This review was limited to a qualitative analysis due to the disparity in reporting and interpreting KAM and the small number of studies in each category. External knee adduction moments were chosen as the primary outcome, as they are still the most commonly used measures of joint loading with an important relationship to disease progression and initiation [10,11]. In recent studies, KAM impulse was investigated and shown to also indicate the onset and progression of knee OA by simultaneously reporting the magnitude and duration of knee loading [67]. However, of the included studies only 6 reported KAM impulse. To broaden the range of this systematic review we chose to focus on peak KAMs rather than including KAM impulse. In addition, alternate measures of joint load are available, including compressive force measured via instrumented joint replacement or by musculoskeletal modeling including muscle activity and forces [68]. Therefore, future research is warranted to determine how the tested gait modification strategies affect knee joint contact forces.

6. Conclusion

This systematic review presents an overview of main factors which can influence the frontal plane knee moment during barefoot gait analysis measurement. Three categories could be defined: gait modifications (speed, trunk lean, in- and out-toeing and step width), individual characteristics (body weight, limb dominance, age) and idiopathic orthopedic deformities (femoral or tibial torsion, genu valgum/varum and foot posture). These factors should be considered when conducting longitudinal studies or multiple gait measurements for therapy planning or for observation purposes. As some studies with the same influencing parameter on KAM found contradictory results, it may be necessary to investigate the effect of these parameters on individuals rather than averaging over all participants.

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Conflict of interest

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7.2. Original Article of Study 2:

Compensatory mechanisms in children with idiopathic lower extremity internal rotational malalignment during walking and running

S. Kimberly Byrnes[1,2], Demir Kunic[1], Roman Rethwilm[1], Harald Böhm[1], Thomas Horstmann[2,3], Chakravarthy U. Dussa[1]

[1]Orthopedic Children's Hospital, Behandlungszentrum Aschau GmbH, Bernauerstr. 18, 83229 Aschau i. Chiemgau, Germany

[2]Department of Sports and Health Sciences, Institute for Conservative and Rehabilitative Orthopedics, Technical University of Munich, Germany

[3]Medical Park St. Hubertus Klinik, Bad Wiessee, Germany.

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Compensatory mechanisms in children with idiopathic lower extremity internal rotational malalignment during walking and running

S. Kimberly Byrnes[a,b,*], Demir Kunic[a], Roman Rethwilm[a], Harald Böhm[a], Thomas Horstmann[b,c], Chakravarthy U. Dussa[a]

[a] Orthopedic Children's Hospital, Behandlungszentrum Aschau GmbH, Bernauerstr. 18, 83229, Aschau im Chiemgau, Germany

[b] Department of Sports and Health Sciences, Institute for Conservative and Rehabilitative Orthopedics, Technical University of Munich, Germany

[c] Medical Park St. Hubertus Klinik, Bad Wiessee, Germany

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ABSTRACT

Background: Noticeable in-toeing gait is present in most children with internal rotational malalignment and often a reason to consult an orthopedic specialist. The risk of tripping may be higher for these patients.

Research Question: The aim of this study was to determine compensatory mechanisms adopted by children with internal rotational deformities to avoid tripping and falling during walking and running.

Methods: Sixty-nine patients between 5–18 years with idiopathic internal rotational malalignment were retrospectively included and subdivided into three groups: 18 patients with internal tibial torsion (ITT), 25 patients with internal femoral torsion (ITF) and 26 patients with both (ITB). Twenty-two typically developing age-matched children (TD) were analyzed for comparison. Three-dimensional gait data were evaluated. ANOVA's on two factors, group (ITT, ITF, ITB, TD) and movement (walking, running) with post-hoc t-tests were used to identify significant differences between groups.

Results: All groups had significantly greater step width than TD during walking ($P \leq .002$) and all torsional groups had significantly greater step width during running ($P \leq .001$). Similarly, all torsional groups showed greater peak ankle dorsiflexion in swing during running than TD ($P \leq .006$). Only the ITT group showed significantly greater external hip rotation than TD. When compared to TD, the ITF and ITB group had a significantly lower hip abduction moment in stance during running, but not for walking ($P \leq .032$).

Significance: Compensatory mechanisms in children with internal rotational deformities were mostly dependent on the location of rotational malalignment. All children with internal rotational malalignment had greater ankle dorsiflexion and greater step width during running. Especially in active patients, this greater ankle dorsiflexion during running may result in overuse of the ankle dorsiflexor muscles, while greater step width may have beneficial effects in normalizing knee adduction moments.

1. Introduction

In-toeing is a noticeable gait deviation in children and therefore a main concern for parents to seek an orthopedic opinion. This gait deviation is caused by different pathological conditions such as increased femoral anteversion and tibial internal torsion, or a combination of both [1]. Besides raising aesthetic concerns, especially for adolescents, in-toeing due to increased femoral torsion can cause problems regarding tripping and falling [2,3]. Furthermore, personal observations during our daily clinical work are in line with Naqvi et al. [4] and have shown that some children with in-toeing gait complain of pain involving the hip or knee during or after running.

Intentional changes in foot rotation in healthy persons during walking and running have been shown to affect the knee adduction moment (KAM) [5–10]. Internal foot rotation is especially known for reducing the first peak of the KAM during walking [6–9] and increasing the second peak of the KAM [5,9]. Therefore, compensatory mechanisms that tend to normalize the foot rotation in in-toeing walkers may result in normal KAM for both peaks. On the other hand, the effects of lower extremity malalignment induced foot progression angle (FPA)

Abbreviations: FPA, foot progression angle; ITB, internal femoral and tibial torsion (both); ITF, internal torsion femur; ITT, internal torsion tibia; OA, osteoarthritis; SD, standard deviation; TD, typical developing children

* Corresponding author at: Gait Laboratory, Orthopaedic Hospital for Children, Behandlungszentrum Aschau GmbH, Bernauerstr, 18, 83229, Aschau im Chiemgau, Germany.

E-mail address: k.byrnes@bz-aschau.de (S.K. Byrnes).

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may differ from those of intentional in-toeing walkers. Additionally, depending on the compensatory mechanism, they may cause non-physiological loading which may lead to overuse and pain in different structures and tissues of the body.

Not all patients with increased femoral anteversion and tibial internal torsion have a noticeable in-toeing gait [11,12]. Rather, in-toeing gait may depend on the degree and level of rotational deformity. For instance, it was shown that femoral anteversion alone has no significant impact on FPA and therefore on in-toeing gait [13]. Another reason that patients may not show an in-toeing gait despite having internal rotational malalignment could be that they tend to develop compensatory mechanisms. For example, the hip may be externally rotated in order to counterbalance the greater internal FPA in children with excessive internal tibial torsion [3]. Similarly, greater pelvic retraction [13], anterior pelvic tilt, knee adduction and hip flexion [14] have been reported in patients with internal femoral torsion during walking. These compensatory mechanisms may provide a safer gait pattern, since they may minimize the risk of tripping over the stance leg. In support of such a concept, compensatory mechanisms in walking have also been reported in children and adults with various pathologies, such as cerebral palsy, hemiplegia, hip dysplasia or osteoarthritis, and are thought to maximize gait efficiency or reduce pain [15].

Although compensatory mechanisms have been commonly reported in children during walking, none of the published studies have evaluated compensatory mechanisms in running to the best of our knowledge. Increased velocity and joint movements during running [16] make it a more demanding and more dynamic task in comparison to walking. With increasing gait speed, sagittal hip, knee and ankle flexion in the swing phase increases [17], which could enhance toe clearance in those with in-toeing. On the contrary, narrower step width associated with running as opposed to walking in healthy persons [18] may make it a more hazardous task for in-toers, as the distance between the limbs during swing decreases and the risk for tripping increased. In support of such a concept, more than half of patients with increased idiopathic femoral anteversion self-report pain as well as greater incidence of tripping and falling during running-related sporting activities [4].

The aim of this study, therefore, was to highlight potential compensatory mechanisms in children and adolescents with in-toeing gait. It was hypothesized that, in comparison to typically developing children, children with increased internal torsion of the lower extremity would demonstrate: 1) greater ankle dorsiflexion, knee flexion and hip flexion, in order to increase toe clearance during mid-swing; 2) larger step width, along with greater external rotation and abduction of the hip and pelvic retraction during stance to avoid tripping over the contralateral leg in the swing phase during walking and running, and; 3) more pronounced compensations during running than walking.

2. Methods

2.1. Participants

A retrospective analysis was done on the data of children and adolescents (5–18 years) who presented to the clinic to seek advice for internal rotation gait between January 2009 and January 2020. As per the hospital protocol all children underwent gait analysis.

A total of 69 consecutive children were included and subdivided into three groups: 18 children (10 female; age: 11.6 ± 2.6 years) with internal tibial torsion (ITT), 25 children (22 female; age: 11.3 ± 3.7 years) with internal femoral torsion (ITF) and 26 children (16 female; age: 10.2 ± 3.4 years) with both, internal femoral and tibial torsion (ITB, Table 1). Rotational deformities were defined as anatomical joint angles that were greater than 1 standard deviation (SD) of those noted for typically developing (TD) children. An internally rotated hip was defined as the hip whose calculated midpoint of rotation was more than 10°. The hip midpoint of rotation was calculated by combining the maximal passive internal rotation (IR) and maximal passive external rotation (ER) and dividing by 2 (IR + ER)/2 [19]. The external rotation was given a negative and the internal rotation a positive value. As a result, the mid-point value indicates the amount and direction of total hip rotation. The internal tibial torsion was defined as a tibial rotation of less than 14°, measured by the relation of the transmalleolar axis to the frontal plane of the thigh. Hip rotations and tibial torsion were measured using a handheld goniometer with the child in a prone position and knees flexed at 90 degrees [1]. The examiner is trained in the clinical orthopedic documentation and has an experience of more than 3 years. Exclusion criteria were: in-toeing gait of non-idiopathic origin, pain induced limitations in running, mental developmental delay, previous surgery on lower extremities, genu varum/valgum, foot deformities, miserable malalignment of legs in all 3 planes, trauma, leg length difference greater than 1 cm, scoliosis and obesity according to the age-dependent body mass index thresholds [20]. Considering the dependence of the legs to one another and the evaluation of the parameter pelvic retraction, only the more affected limb of each child was included. Twenty-two typically developing (TD) children and adolescents (11 female; age: 10.4 ± 2.5 years) were analyzed for comparison. All parents and children provided general consent.

Table 1
Mean (standard deviation) anthropometrics and clinical values of femoral and tibial torsion (positive values indicate internal, negative indicate external position) for the more affected leg of children with internal deformity and TD;

Parameter	Group			
	ITF (n = 25)	ITB (n = 26)	ITT (n = 18)	TD (n = 22)
Hip rotation midpoint [°]	30.9(8.6)	25.9(9.08)	5.69(5.61)	3.26(6.8)
Tibial torsion [°]	−20.9(3.65)	−8.62(3.84)	−7.22(4.58)	−22.1(8.66)
Age [years]	11.3(3.7)	10.2(3.36)	11.6(2.62)	10.4(2.46)
Body height [cm]	150(18.4)	145(14.6)	153(13.2)	143(14.6)
Body weight [kg]	41.8(15.9)	40.3(14)	46.3(14.9)	36(11.2)
Body mass index [kg/m^2]	17.8(3.34)	18.5(3.18)	19.3(3.64)	17.2(2.26)

SD – standard deviation; ITF – patient group with increased femoral anteversion; ITB – patient group with increased femoral anteversion and tibial internal torsion; ITT – patient group with tibial internal torsion; TD – typically developing children.

2.2. Data collection and evaluation

Kinematic and kinetic data were collected using an 8-camera system (Vicon Motion Systems Ltd., Oxford, UK) and two force plates (AMTI, Watertown, MA, USA). In addition to the standard Vicon Plug-in-Gait marker set, medial ankle and knee markers were used during the static trial to improve the accuracy of the calculation of joint rotations in the transverse plane [21].

Participants were asked to walk and run barefoot at their own comfortable speed up and down a 13-m walkway until five gait cycles of walking and running with valid foot strikes on the force platform were captured. Additionally, all participants underwent a thorough clinical examination subsequent to gait analysis and patients were asked to state any existing pain.

The following gait parameters were inspected for the appearance or result of compensatory mechanisms: step width, minimum toe clearance during mid-swing (between 45 %–55 % of swing phase [22]), peak pelvic retraction during the stance phase of gait, peak hip, knee and ankle flexion in swing, mean FPA in stance, mean internal hip abduction moment in stance and mean internal knee adduction moment in the stance phase of gait (Table 2). The type of foot-strike running pattern was categorized based on the mean sole angle at initial contact which is the angle between the long axis of the foot to the ground (forefoot > 0°, heelstrike ≤ 0°). To account for differences in leg length

Table 2
Mean values (standard deviation) of spatio-temporal parameters, peak segment and joint moments for walking and running. Significant results between patients and TD were marked in bold after controlling for false discoveries according to the Benjamini-Hochberg procedure and the critical p-value of ≤ .036. Positive values indicate internal, negative indicate external movement for parameters in the transversal plane.

	walk				run			
Parameter	*ITF*	*ITB*	*ITT*	*TD*	*ITF*	*ITB*	*ITT*	*TD*
Velocity [m/s]	1.3(0.15)	1.3(0.12)	1.29(0.15)	1.27(0.13)	2.8(0.41)	2.68(0.4)	2.62(0.5)	2.76(0.41)
Nondimensional velocity	0.47(0.05)	0.48(0.05)	0.46(0.05)	0.47(0.06)	1.01(0.17)	0.99(0.18)	0.93(0.18)	1.03(0.2)
Nondimensional cadence	0.60(0.04)	0.60(0.03)	0.59(0.05)	0.59(0.04)	0.90(0.05)	0.89(0.07)	0.87(0.07)	0.87(0.07)
Step width [cm]	**8.98(1.76)**	**11.2(1.94)**	**10.6(2.26)**	7.39(1.54)	**6.5(2.35)**	**8.78(3.06)**	**7.68(3.64)**	4.37(1.67)
Step length [cm]	78.6(7.13)	78.2(5.45)	77.4(6.28)	78.9(6.95)	113(16.8)	112(17.7)	106(15.8)	118(19.2)
Minimum toe clearance swing [cm]	6.32(1.05)	6.57(1.29)	6.84(0.946)	6.35(1.06)	11.1(4.74)	11.2(4.37)	10.4(3.78)	13.9(5.34)
Peak pelvic retraction stance [cm]	**−10.2(4.43)**	**−10.6(4.84)**	−7.85(3.91)	−6.79(3.98)	−5.71(4.99)	−7.39(3.58)	−6.49(3.82)	−7.01(3.34)
Peak hip flexion swing [°]	36.3(6.82)	38.7(6.92)	**31.8(6.38)**	36.1(5.25)	48.8(6.72)	51.7(8.91)	**42.4(7.39)**	49.5(8.52)
Peak knee flexion swing [°]	57.9(5.11)	59.3(3.95)	57.4(3.87)	59.5(2.48)	**78.4(11.1)**	81(10.6)	**77.1(7.66)**	86.2(11.6)
Peak ankle dorsiflexion swing [°]	5.56(3.11)	6.96(3.22)	6.58(3.86)	5.03(3.62)	**10.7(5.69)**	**11(7.07)**	**12.7(5.97)**	5.5(5.76)
Mean hip rotation stance [°]	**12.9(5.68)**	5.74(8.67)	**0.27(7.19)**	5.65(7.24)	**13.5(5.35)**	7.89(6.89)	**2.01(7.5)**	9.16(6.78)
Mean foot progression stance [°]	**4.28(5.88)**	**8.03(5.65)**	**8.12(4.05)**	−5.72(5.9)	**1.78(6.71)**	**7.84(6.58)**	**6.6(5.43)**	−7.51(7.09)
Mean hip abduction moment stance [Nm/kg]	0.41(0.11)	0.41(0.1)	0.46(0.13)	0.42(0.08)	**0.62(0.14)**	**0.55(0.14)**	0.65(0.23)	0.71(0.16)
1st peak knee adduction moment stance [Nm/kg]	0.42(0.12)	0.43(0.13)	0.46 (0.14)	0.41(0.12)	0.80(0.27)	0.93(0.32)	**1.11(0.36)**	0.85(0.36)
2nd peak knee adduction moment stance [Nm/kg]	0.31(0.13)	0.30(0.09)	**0.38(0.11)**	0.29(0.1)				

ITF – patient group with increased femoral anteversion; ITB – patient group with increased femoral anteversion and tibial internal torsion; ITT – patient group with tibial internal torsion; TD – typically developing children.

at different ages, non-dimensional velocity and cadence were calculated and used for statistical evaluation [23]. Mean values for gait parameters were calculated as the average over the stance phase, while peak values reflect the maximum or minimum in stance or swing phase of each gait cycle averaged over 5 gait cycles for each child.

For each dependent variable a two-factor (group, movement) analysis of variance (ANOVA) was used for statistical evaluation between groups (ITF, ITT, ITB and TD). Post-hoc t-tests were performed to detect significant speed and interaction effects (Table 3). The Benjamini-Hochberg procedure was performed to control for false discovery rates. A false discovery rate of 10 % was used.

3. Results

In total, 30 % of children and 45 % of TD adopted a forefoot footstrike pattern during running. The distribution of the forefoot footstrike pattern was 32 %, 33 % and 27 % for the ITF, ITT, and ITB groups, respectively. Pain in the hip, knee or shin during or after running or sporting activities was reported by 7 children of the ITF group and 4 children of the ITT group but none in the ITB group. The critical p-value after controlling for false discoveries according to Benjamini-Hochberg was ≤ 0.058 for the ANOVA and ≤0.036 for the post-hoc tests. All evaluated parameters changed significantly between walking and running in all groups. Significant differences between internal rotational deformity groups and TD are highlighted below.

3.1. Spatial-temporal parameters

Self-selected walking speed was not significantly different between groups (Table 2). TDs walked at 1.3 m/s (SD = 0.1) and ran at 2.8 m/s (SD = 0.4). Step width was significantly greater for all groups compared to TD during walking ($P \leq .002$) and during running ($P \leq .001$).

3.2. Kinematics

Minimum toe clearance at mid-swing did not differ between all groups compared to TD (Fig. 1). Peak pelvic retraction in stance showed an interaction effect and was greater in ITF and ITB groups only during walking (P = .008 and P = .005, respectively, Fig. 2). The ITT group had less peak hip flexion in swing during walking and running compared to TD (P = .024 and P = .008, respectively). The ITF and ITT group had less peak knee flexion during running than TD (P = .022 and P = .007, respectively). For peak ankle dorsiflexion, there were significant differences between internal rotational deformity groups and TD only during running (Fig. 3). Children with internal rotational deformity had greater peak ankle dorsiflexion in the swing phase compared to TD (ITF: P = .003; ITB: P = .006; ITT: $P < .001$). ITF, ITB and ITT had a significant increase in peak ankle dorsiflexion from walking to running, whereas the peak ankle dorsiflexion in the TD group stayed the same. The mean hip rotation in stance during walking was significantly different between the ITF and TD ($P < .001$) and the ITT and TD groups (P = .024). While the ITF group had significantly greater hip internal rotation, the ITT group showed a significantly greater hip external rotation. The mean foot progression angle showed a significant group and speed effect. All patient groups had significant greater internal FPA compared to TD during walking and running ($P < .001$).

3.3. Kinetics

During running, the ITF and ITB group had a lower mean hip abduction moment during stance than TD (P = .032 and P = .001, respectively). The second peak of the KAM during walking and the peak KAM during running is significantly greater for the ITT group (P = .015 and P = .032, respectively).

4. Discussion

The hypotheses that children and adolescents with internal rotational deformities adopt compensatory mechanisms in gait could partly be confirmed. Although compensatory mechanisms were evident in patients with internal rotational deformities, they were mostly dependent on the location of the torsional deformity.

4.1. Compensatory mechanisms during walking and running

Our data showed that in-toeing gait was more pronounced in the ITT (100 %) and ITB (92 %) than ITF group (76 %). This finding is comparable to that reported by Radler et al. [13]. As anticipated, the ITF group also showed a more internally rotated hip than the TD group. We assume that ITF children do not fully compensate hip internal rotation with hip external rotation despite passive external rotation ability of on average 16°. The FPA was less pronounced which may make it unnecessary to counterbalance in-toeing gait. Even though the ITB group had similar passive midpoint of hip rotation as the ITF group, ITB were able to present a dynamic hip rotation similar to TD. Hence

Table 3
ANOVA and post-hoc t-tests results for spatio-temporal parameters, peak segment and mean joint moments. Comparison between groups and walking/running are shown. Significant results are marked in bold according to the Benjamini-Hochberg procedure with p ≤ .058 for the ANOVA and p ≤ .036 for the post hoc tests.

	ANOVA			Post hoc *t* -test walk						Post hoc *t* -test run						Post hoc *t* -test walk - run			
Parameter	*Group*	*Speed*	*IA*	*F-TD*	*B-TD*	*T-TD*	*F-T*	*F-B*	*B-T*	*F-TD*	*B-TD*	*T-TD*	*F-T*	*F-B*	*B-T*	*F-F*	*B-B*	*T-T*	*TD-TD*
Velocity	0.626	**< .001**	0.464													**< .001**	**< .001**	**< .001**	**< .001**
Nondimensional velocity	0.406	**< .001**	0.406													**< .001**	**< .001**	**< .001**	**< .001**
Nondimensional cadence	0.415	**< .001**	0.536													**< .001**	**< .001**	**< .001**	**< .001**
Step width	**< .001**	**< .001**	0.757	**0.002**	**< .001**	**< .001**	**0.011**	**< .001**	0.4	**0.001**	**< .001**	**0.001**	0.203	**0.005**	0.286	**< .001**	**< .001**	**< .001**	**< .001**
Step length	0.243	**< .001**	0.192													**< .001**	**< .001**	**0.001**	**< .001**
Minimum toe clearance swing	0.725	**< .001**	0.491													**< .001**	**0.002**	0.253	0.813
Peak pelvis retraction stance	0.174	**< .001**	**0.003**	**0.008**	**0.005**	0.402	0.075	0.756	0.049	0.305	0.709	0.65	0.578	0.172	0.432	**< .001**	**0.002**	0.253	0.813
Peak hip flexion swing	**0.001**	**< .001**	0.564	0.906	0.157	**0.024**	**0.033**	0.222	**0.002**	0.731	0.399	**0.008**	0.005	0.193	**0.001**	**< .001**	**< .001**	**< .001**	**< .001**
Peak knee flexion swing	**0.021**	**< .001**	0.097	0.17	0.826	0.045	0.767	0.259	0.125	**0.022**	0.111	**0.007**	0.688	0.39	0.195	**< .001**	**< .001**	**< .001**	**< .001**
Peak ankle dorsiflexion swing	**0.007**	**< .001**	**0.003**	0.594	0.057	0.2	0.345	0.122	0.724	**0.003**	**0.006**	**< .001**	0.257	0.876	0.387	**< .001**	**0.002**	**< .001**	0.675
Mean hip rotation stance	**< .001**	**< .001**	**0.057**	**< .001**	0.967	**0.024**	**< .001**	**0.001**	**0.033**	**0.017**	0.524	**0.003**	**< .001**	**0.002**	**0.01**	0.343	**0.006**	**0.021**	**< .001**
Mean foot progression stance	**< .001**	**0.003**	0.359	**< .001**	**< .001**	**< .001**	**0.022**	**0.024**	0.954	**< .001**	**< .001**	**< .001**	**0.016**	**0.002**	0.512	**0.003**	0.851	0.252	0.081
Mean hip abduction moment stance	0.073	**< .001**	**0.001**	0.822	0.668	0.178	0.164	0.876	0.098	**0.032**	**0.001**	0.309	0.562	0.112	0.091	**< .001**	**< .001**	**< .001**	**< .001**
1st peak knee adduction moment stance	**0.058**	**< .001**	**0.01**	0.752	0.547	0.226	0.322	0.753	0.491	0.62	0.425	**0.032**	0.003	0.138	0.096	**< .001**	**< .001**	**< .001**	**< .001**
2nd peak knee adduction moment stance	**0.001**			0.569	0.84	**0.015**	0.096	0.655	**0.012**										

IA – Interaction effect; F – femoral anteversion; B – both (tibial and femoral internal torsion); T – tibial internal torsion; TD – typically developing children;

external hip rotation occurred to offset femoral internal torsion and compensate in-toeing gait; thus resulting in normal hip rotation. However, mean step width did not differ between ITT and ITB groups. The normal hip rotation, the slightly less pronounced tibial torsion and thus the smaller FPA in the ITB group may result in a similar step width to ITT.

The ITT group was the only group that showed a more externally rotated hip than TD. The external rotation of the hip in the stance phase may be the main clinically relevant compensatory mechanism against the internal torsion of the tibia [3,13]. An excessive external hip rotation is reported to cause patellofemoral knee pain in children [24]. Hence, the children in this group may be predisposed to patella-femoral pain over time. Furthermore, greater KAM in the ITT group could influence the development of pain and long term knee joint health [3]. However, during walking only the 2nd peak of KAM was significantly higher compared to the KAM of TD. The clinical implication of this is still not known. A greater 1 st peak of KAM has been associated with degenerative knee joint disease in the literature [25,26]. Both peaks of the KAM in ITF and ITB groups were similar to those in the TD group. A wider step width may have an influence on normalizing the KAM in these children [8,9].

All internal rotational deformity groups in this study had a greater step width than TD children. Greater step width affords more space for the toes of the swing limb to pass by the stance leg, and a lower risk of tripping. This may be especially important for running tasks where tripping occurs more frequently and possibly with greater consequences.

4.2. Compensatory mechanisms only during walking

The pelvis was retracted for the ITF and ITB group, which may contribute to toe clearance during the swing phase of gait. This finding is in accordance to previous research, which showed increased pelvic retraction to compensate the increased anteversion [13]. Only the two groups of children that were affected at the hip in this study showed this compensatory mechanism.

4.3. Compensatory mechanisms only during running

Both ITF and ITB groups had a significantly lower hip abduction moment (with the abduction moment being the lowest in the ITB group) in stance than TD. A previous study identified the internal rotation of the hip as a cause for lower abduction moment [27]. This was observed for the ITF. However, the ITB group showed the lower abduction moment in spite of normal hip rotation. In contrast to the ITF group the ITB group showed greater step width, which could have played a role in lowering the hip abduction moment [24]. Moreover, trunk lean is also known to influence the hip moments in the frontal plane [29], however, this was not investigated in this study.

All in-toeing children in the current study had greater peak ankle dorsiflexion in swing than TD children. The greater ankle dorsiflexion in the swing phase ($5-6^{\circ}$) is likely to be clinically meaningful and may help to avoid inadvertent contact of the swing limb to the stance leg by achieving more toe clearance. Possibly due to the greater dorsiflexion in swing the hip and knee may be less flexed. To gain greater ankle dorsiflexion more work needs to be generated by the anterior muscles of the lower leg, such as the tibialis anterior muscle. This has the potential to result in overuse injuries of the muscle tendon unit, especially in active patients [30].

Outcomes of untreated internal rotational malalignment is not well understood and may not be only aesthetic, and could lead to orthopedic problems [12]. Internal rotational deformities could potentially influence the development of knee or hip osteoarthritis. However, the association is debated in the literature. An *in vitro* study found that torsional deformities of the lower extremities could not predict the development of knee or hip osteoarthritis [31]. In contrast, a clinical

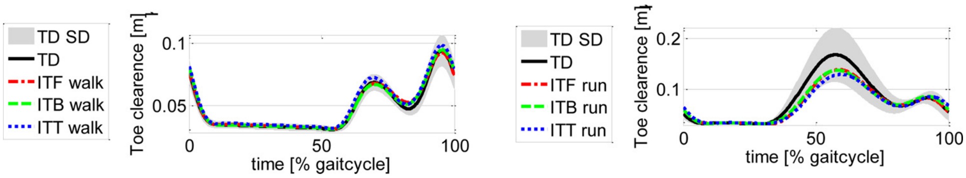

Fig. 1. Toe clearance during walking (left) and running (right) of patients with internal femoral torsion (ITF), internal tibial torsion (ITT) and combined internal femoral and tibial torsion (ITB) and in typical developing children (TD).

study has shown that patients with knee osteoarthritis have less external tibial torsion than those who do not [32], and a biomechanical study has shown greater knee loading in patients with in-toeing gait, which did not normalize after surgical treatment [3].

One potential limitation of this study was that classification of patients was based on clinical assessment rather than medical imaging, because not all patients had recently undergone magnetic resonance imaging. Furthermore, studies have shown sex differences in adult gait kinematics, particularly in the frontal and transverse plane [33]. While the sample size of the current study prohibits meaningful sub-analysis, it is possible that compensatory mechanisms may be moderated by sex differences. However, the hip midpoint of rotation between girls and boys in our norm cohort was not significantly different. Detailed information about the results of girls and boys are presented in supplementary data tables. Another potential limitation of this study is that the running distance of 13 m may be considered too short for the children to achieve their typical running pattern. The typical definition of running gait (*i.e.* single support phase) however was present in all participants and running velocities are representative of those reported elsewhere for similarly aged children [34,35]. Nonetheless it is assumed that running patterns may differ between laboratory and outdoor settings.

5. Conclusion

Children and adolescents with idiopathic internal rotational malalignment of the lower extremity display different compensatory mechanisms depending on the location of the deformity. During walking, we found external hip rotation (ITT) and pelvic retraction (ITF and ITB) and greater step width for all in-toeing children. During running, we found a lower hip abduction moment (ITF and ITB) and greater step width and greater ankle dorsiflexion in all in-toeing groups. Clinicians should be aware of compensatory mechanisms as they may lead to pain and in which case surgery may be indicated. Furthermore, compensatory mechanisms seem to have positive effects on normalizing the knee adduction moment in ITF and ITB and negative effects in ITT, but it is still unknown if these effects disappear or result in non-physiological knee loading conditions after derotation surgery.

Declaration of Competing Interest

All authors do not have any financial and personal relationships with other people or organizations that inappropriately influence the work performed. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

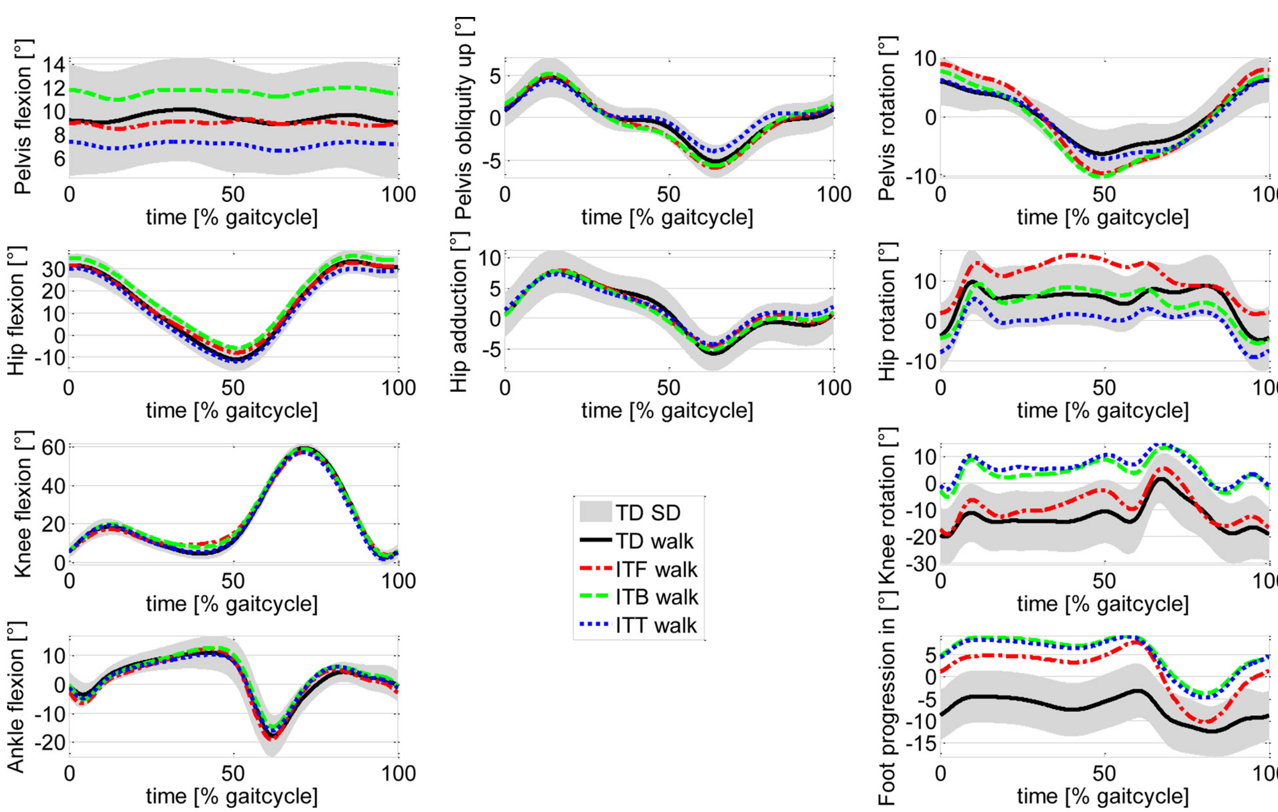

Fig. 2. Kinematic time curves in the sagittal (left), frontal (middle) and transverse plane (right) of patients with internal femoral torsion (ITF), internal tibial torsion (ITT) and combined internal femoral and tibial torsion (ITB) and in typical developing children (TD) during walking. Negative values indicate internal rotation, while positive values indicate external rotation for the transverse plane.

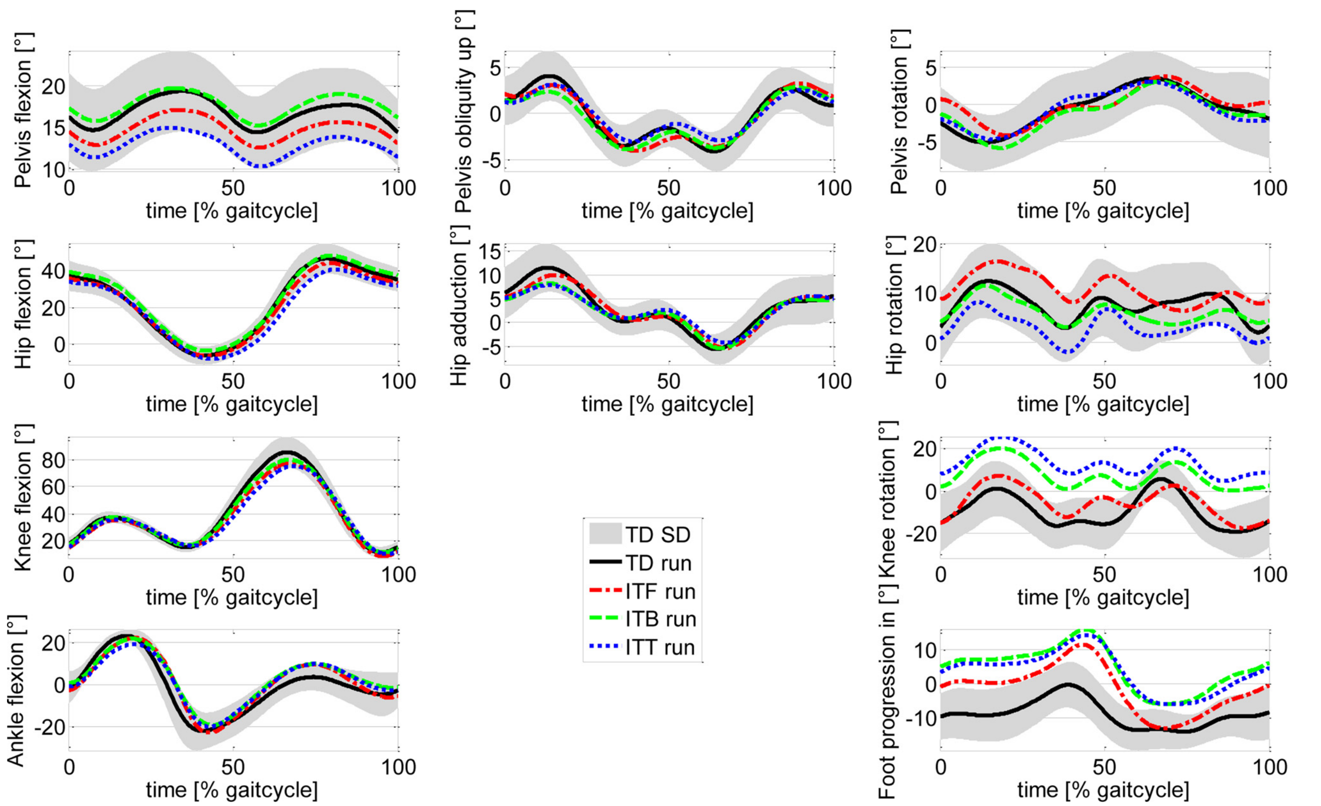

Fig. 3. Kinematic time curves in the sagittal (left), frontal (middle) and transverse plane (right) of patients with internal femoral torsion (ITF), internal tibial torsion (ITT) and combined internal femoral and tibial torsion (ITB) and in typical developing children (TD) during running. Negative values indicate internal rotation for the transverse plane.

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Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.gaitpost.2020.03.015.

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7.3. Original Article of Study 3:

Effects of idiopathic flatfoot deformity on knee adduction moments during walking

S. Kimberly Byrnes[1,2], Scott Wearing[2,3], Harald Böhm[1], Chakravarthy U. Dussa[1], Thomas Horstmann[2,4]

[1]Orthopedic Children's Hospital, Behandlungszentrum Aschau GmbH, Germany

[2]Department of Sports and Health Sciences, Institute for Conservative and Rehabilitative Orthopedics, Technical University of Munich, Germany

[3]Institute of Health and Biomedical Innovation, Queensland University of Technology, Australia

[4]Medical Park St. Hubertus Klinik, Bad Wiessee, Germany.

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Effects of idiopathic flatfoot deformity on knee adduction moments during walking

S. Kimberly Byrnes [a,b,*], Scott Wearing [c,d], Harald Böhm [a], Chakravarthy U. Dussa [a], Thomas Horstmann [b,d]

[a] *Orthopedic Children's Hospital, Behandlungszentrum Aschau GmbH, Germany*
[b] *Department of Sports and Health Sciences, Institute for Conservative and Rehabilitative Orthopedics, Technical University of Munich, Germany*
[c] *Institute of Health and Biomedical Innovation, Queensland University of Technology, Australia*
[d] *Medical Park St. Hubertus Klinik, Bad Wiessee, Germany*

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ABSTRACT

Introduction: Flatfoot deformity is commonly characterized by a subtalar valgus, a low medial longitudinal arch, and abduction of the forefoot. Although flatfoot deformity has been associated with lower first (KAM1) and second (KAM2) peak knee adduction moments during walking, the biomechanical connection remains unknown.
Research question: We hypothesized that hindfoot eversion, lateral calcaneal shift correlate with KAM1 and forefoot abduction and arch height with KAM2, due to the lateralization of the ground reaction force vector resulting from shifted heel and forefoot in flatfoot deformity.
Methods: Gait data from 103 children with flatfoot deformity who underwent three-dimensional gait analysis with the Oxford Foot Model were retrospectively included. Children with knee varus/valgus, in- and out-toeing were excluded. Fifteen healthy children with a rectus foot type were also collected from the database. Lateral calcaneal shift was defined as the distance between the projection of the ankle joint center onto the calcaneal axis and the midpoint of the calcaneal axis formed by the medial and lateral calcaneal markers. A subgroup of children with idiopathic flatfoot deformity that had received corrective surgery was also identified. Statistical analysis included Pearson's correlations and independent and paired t-tests ($\alpha < .05$).
Results: When compared to a norm cohort, flatfooted children had significant lower KAM1 and KAM2 (t-test, $P < .001$). Lateral calcaneal shift correlated with KAM1 and KAM2 ($r = 0.42$, $p < .001$ and $r = 0.32$, $P < .001$, respectively). Arch height correlated with KAM2 ($r = 0.23$, $p = 0.017$). KAM1 and KAM2 normalized after surgery and the change in KAM1 correlated with the change in lateral calcaneal shift for children who underwent corrective surgery.
Significance: Lateral calcaneal shift explains the reduction of KAM1 by lateralization of the point of force application in flatfooted children. It is recommended to consider the lateral calcaneal shift when investigating KAM in gait analysis research.

1. Introduction

Flatfoot deformity is a common condition in children and adolescents [1], which is characterized by a low medial longitudinal arch (MLA), hindfoot valgus and forefoot abduction and supination. The primary pathology of flatfoot deformity involves decentering of the talo-navicular joint resulting in subluxation of the subtalar joint. Hindfoot valgus occurs either secondary to eversion of the heel or due to a lateral shift of the heel or both [2].

Some children with flatfoot deformity are symptomatic and report pain, not only involving the foot but also the knee, hip and back [3,4]. Other children may not report pain but their foot function might still be compromised [4]. Suboptimal foot function and pain may indicate alterations in gait mechanics which can cause an imbalance of

Abbreviations: FF, flatfoot; KAM, knee adduction moment; KAM1, first peak of knee adduction moment; KAM2, second peak of knee adduction moment; LCA, lateral calcaneal; MLA, medial longitudinal arch; NF, normal rectus foot type; OFM, Oxford Foot Model; PiG, Plug-In-Gait Model; STL, *Sustentaculum Tali*.

* Corresponding author at: Gait Laboratory, Orthopedic Hospital for Children, Behandlungszentrum Aschau GmbH, Bernauerstr. 18, 83229 Aschau i. Chiemgau, Germany.

E-mail address: k.byrnes@bz-aschau.de (S.K. Byrnes).

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load-distribution by unloading certain joints and segments consequently resulting in overuse in others.

Gait kinematics and kinetics of the distal and proximal joints of the lower limb have been well described in children with flatfeet [3,5,6]. In particular, flatfoot deformity in children has been associated with changes in the knee adduction moment (KAM) during walking [3]. For instance, Kothari et al. [3] reported a reduction in the second peak of the knee adduction moment (KAM2) in children with flatfoot deformity compared to children with a rectus foot type [3]. However, flatfoot deformity leads to various gait deviations [3,5,6] presumably because the subcomponents of the deformity show dominance in different planes of the foot [7]. It is still not well known how these subcomponents influence the knee adduction moment. Given that changes in KAM are indicative of knee joint loading [8] and have been associated with knee osteoarthritis in large cohort studies [9,10], it is important to further evaluate KAM in children with flatfeet.

The aim of this study, therefore, was to investigate the impact of flatfoot deformity on KAM in children during barefoot walking and whether the predominant plane of deformity (transverse, frontal or sagittal) is related to peak knee adduction moments. We specifically hypothesized that children with flatfoot deformity would have lower KAM1 and KAM2 compared to children with a normal rectus foot type (NF) and that KAM1 and KAM2 would normalize following corrective surgery for flatfoot. Biomechanically, it is possible that a more lateral position of the calcaneus (frontal plane) at heel strike lowers KAM during the loading response phase of walking (KAM1), due to a more lateral point of heel impact at initial contact. A lowering of KAM in terminal stance phase of walking (KAM2), in contrast, would be expected with increased forefoot abduction (transverse plane) due to a more lateral position of the forefoot and subsequent lateral shift of the ground reaction force vector. A lowered MLA (sagittal plane) on the other hand has previously been associated with lower KAM2 [3]. Therefore, we hypothesized that hindfoot eversion and lateral calcaneal shift would be associated with a lower KAM1 during walking in children with flatfoot deformity, while forefoot abduction and lowered MLA would result in a lower KAM2 compared to NF.

2. Methods

This retrospective cohort study compared lower limb kinematics and kinetics in children with flatfoot deformity during walking with those of a reference group of similarly aged healthy children with a normal rectus foot type. Gait analysis was repeated in a subgroup of children with flatfoot deformity that underwent corrective surgery to further elucidate whether changes in flatfoot deformity were related to changes in KAM. Written general consent for future use of clinical data was obtained from all patients at the time of treatment. Given the retrospective nature of the study, the Bavarian Medical Council granted the project an exemption from ethical review.

2.1. Study cohort

One hundred and three children and adolescents, aged 6–17 years, that presented to a children's hospital between 2011 and 2019 with flatfoot deformity and that underwent gait analysis for orthopedic assessment were included in this study (Table 1). In all cases idiopathic flatfoot deformity was diagnosed by pediatric orthopedists. Excluded were orthopedic deformities and related gait disorders that are known to affect KAM: Children with out-toeing or in-toeing gait (as defined by a foot progression angle of the mean plus 1 standard deviation outside the range of -19.1° – 4.7° from healthy children) secondary to external rotational deformities of the hip (-4.2°) or tibia (-28.8°) were excluded [11–13]. Similarly, children with diagnosed knee varus/valgus deformity by the pediatric orthopedist [14], neurological disorders, previous orthopedic surgery of the lower extremities or obesity, defined by age-dependent body mass index threshold were excluded.

Table 1
Anthropometric data for children with flatfeet (FF) and children with a normal rectus foot type (NF). Significant differences between groups are marked with *.

	FF (n = 103)	NF (n = 15)	P	FFpre (n = 19)	FFpost (n = 19)	P
Female/male	39/64	9/6		9/10	9/10	
Age [years]	11.7 (2.3)	12.9 (3.4)	0.060	11.3 (1.9)	13.3 (2.5)	0.009*
Body height [cm]	154 (13)	154 (18)	0.974	156 (15.1)	164(14)	0.069
Body weight [kg]	44.4 (11.4)	48.7 (14.7)	0.198	45.1 (10.9)	54.6 (13.6)	0.023*
BMI [kg/mˆ2]	18.4 (2.6)	19.8 (2.9)	0.050*	18.4 (2.2)	20(3.12)	0.083

FFpre: Subgroup of children with flatfoot deformity before surgical correction. – FFpost: after surgical correction.

A subgroup of 19 children that underwent corrective surgery for flatfoot deformity was identified to evaluate whether a change in subcomponents of flatfoot deformity were associated with a change in KAM. Surgical correction involved arthroreisis with (n = 3) and without (n = 16) medializing calcaneal osteotomy. In nine children, additional muscle (M. Gastrocnemius) or tendon (Achilles tendon) lengthening was performed.

Reference values were also collected from 15 healthy children that presented with normal lower limb alignment and a normal rectus foot type (Table 1).

2.2. Procedure

Kinematic data was collected by an infrared-based three-dimensional motion analysis system (8-camera system, 200 Hz, Vicon Motion Systems Ltd., Oxford, UK). Two standard force plates (1000 Hz, AMTI, Watertown, MA, USA) arranged in staggered configuration and integrated into a 13-m walkway captured three-dimensional ground reaction forces during walking. Reflective markers were placed on specific anatomical landmarks according to a modified Plug-In-Gait Model (PiG) [15] and the Oxford Foot Model (OFM) [16].

Data collection was preceded by a 3-minute familiarization period during which the starting position for each child was adjusted such that their foot would strike the force platforms without notable alterations to their gait pattern. Children walked barefoot at their preferred speed. Five gait trials that included valid foot strikes on the force platforms were recorded. Marker trajectory noise was filtered using a quintic spline (Woltring) algorithm with the mean square error setting of 12. The OFM and PiG model were run with the Vicon Nexus Software version 1.8 for older and 2.8 for recent collected data.

2.3. Parameters

The internal knee adduction moment was calculated using an inverse dynamics approach [17] and normalized to body mass. Walking velocity and step width were normalized to leg length [18]. The peak knee adduction moments during the first (KAM1) and second (KAM2) half of the stance phase were identified. Hindfoot eversion, lateral calcaneal shift, forefoot abduction and MLA height were subsequently investigated at the exact instance of KAM1 and KAM2 occurrences. The OFM provides detailed information about hindfoot eversion, MLA height and forefoot abduction [16]. Although hindfoot eversion in relation to the axis of the tibia is often used in clinical settings to assess the magnitude and progression of flatfoot deformity, calcaneal shift is an additional parameter that we developed to quantify flatfoot deformity (Fig. 1). Calcaneal shift can be readily derived from the OFM marker set but is not a default output parameter. Calcaneal shift is calculated by projecting

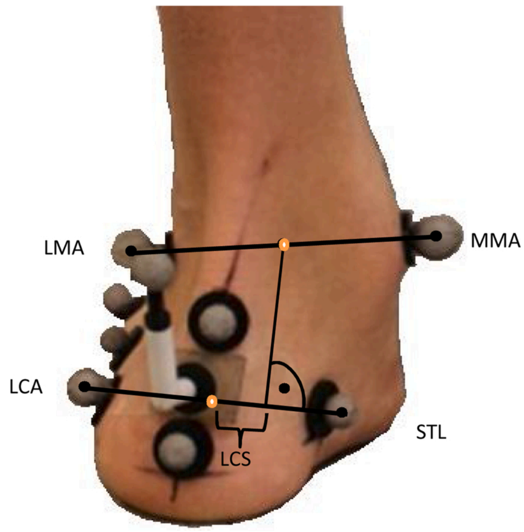

Fig. 1. Graphical presentation of the calculation of the lateral calcaneal shift. Lateral calcaneal shift (LCS) is indicated as percentage of the calcaneal width which is represented by the distance between the LCA and STL markers. MMA: medial malleolus. – LMA: lateral malleolus. – LCA: lateral calcaneus. – STL: sustentaculum tali.

the center point of the medial and lateral ankle joint markers to line connecting the medial sustentaculum tali (STL) and lateral calcaneal (LCA) markers (Fig. 1). The distance from the projected point to the center of the STL and LCA markers defines calcaneal shift and is normalized to the calcaneal width (distance STL to LCA). Flatfoot deformity is typically characterized by a hindfoot valgus, which can result from a hinged lateral rotation of the calcaneus under the talus (described by the parameter hindfoot eversion) or from a lateral translation of the calcaneus under the talus (described by the parameter lateral calcaneal shift) [2]. Mean hindfoot eversion, lateral calcaneal shift, forefoot abduction and MLA height at the time point of KAM1 and KAM2 were subsequently calculated over the five walking trials.

2.4. Statistical analysis

Statistical analysis was performed with MATLAB student version R2020a (The Mathworks Inc., Natick, USA). Independent t-tests were used to evaluate potential differences in KAM, hindfoot eversion, lateral calcaneal shift, forefoot abduction and arch height in FF and NF children. Similarly, differences in KAM, hindfoot eversion, lateral calcaneal shift, forefoot abduction and arch height with corrective surgery were investigated using paired tests ($\alpha < .05$). Pearson's correlations were used to explore potential relationships among KAM1 and KAM2, hindfoot eversion, lateral calcaneal shift, forefoot abduction and arch height. Correlation coefficients between 0.10 and 0.39 were considered weak, 0.40 to 0.69 as moderate, 0.70 to 0.89 as strong and greater than 0.90–1.00 as very strong [19].

3. Results

There was no statistically significant difference in mean age, body height or body weight of FF and NF (Table 1).

3.1. Peak KAM in children with and without flatfeet

Preferred walking speed did not differ significantly between FF and NF (Table 2). However, step width was significantly greater (+3 cm) in FF than NF children ($p < .001$). Similarly, KAM1 and KAM2 were significantly lower (≈ 30 %) in FF than NF children (Fig. 2; $p < .05$).

3.2. Relationship between KAM and foot posture

As shown in Fig. 3, only lateral calcaneal shift in FF was significantly correlated with both peaks in KAM during walking, demonstrating a moderate to weak correlation with KAM1 ($r_{KAM1} = 0.42$, $p < .001$) and KAM2 ($r_{KAM2} = 0.32$, $p < .001$), respectively. Children with greater lateral calcaneal shift had lower KAM peaks during walking. MLA height showed a weak correlation but only with KAM2 ($r = 0.23$, $p = 0.017$).

Similar relationships were evident in NF. Lateral calcaneal shift was moderately correlated with KAM1 ($r = .70$, $p = 0.004$) during walking and, although not statistically significant, also tended to be correlated with KAM2 ($r = 0.50$, $p = 0.06$). As in children with FF, MLA height was moderately correlated with KAM2 ($r = .60$, $p = 0.02$), in NF children.

3.3. KAM in surgically corrected feet

In total, 19 of the 103 children that presented with flatfeet received

Table 2
Kinematic and kinetic data for children with flatfeet (FF) and children with a normal rectus foot type (NF). Significant differences between groups are marked with *. Non-dimensional parameters were normalized to leg length.

	FF (n = 103)	NF (n = 15)	P	FFpre (n = 19)	FFpost (n = 19)	P
Time after surgery [months]	–	–		–	14.8(9.9)	
Velocity [m/s]	1.3(0.2)	1.3(0.1)	0.731	1.3(0.2)	1.3(0.2)	0.452
Velocity non dimensional	0.47(0.06)	0.47(0.04)	0.522	0.45(0.05)	0.45(0.07)	0.966
Step width [cm]	10.5 (2.5)	7.5(1.9)	<.001*	10.4(2.3)	10.1(1.9)	0.486
Step width non-dimensional	0.13 (0.07)	0.1 (0.03)	0.033*	0.16 (0.14)	0.12 (0.02)	0.198
KAM1 [Nm/kg]	0.37(0.15)	0.54(0.18)	<.001*	0.34(0.13)	0.44(0.16)	0.020*
KAM2 [Nm/kg]	0.26(0.12)	0.37(0.10)	0.001*	0.21(0.10)	0.32(0.11)	<.001*
Mean vertical ground reaction force in stance [% body weight]	84.0(3.4)	83.5(3.6)	0.605	84.2(2.0)	82.9(1.8)	0.046*
Mean knee adduction angle in stance [°]	−1.7(4.0)	−1.4(2.0)	0.794	−2.2(2.9)	−3.2(2.8)	0.263
Hindfoot eversion T1 [°]	−3.7(4.9)	2.7(5.0)	<.001*	−4.4(5.5)	−1.6(6.5)	0.132
Hindfoot eversion T2 [°]	4.6(5.6)	9.3(7.1)	0.004*	6.4(7.3)	7.8(5.3)	0.451
Lateral calcaneal shift T1 [% calcaneal width]	−26.7(11.0)	−9.8(7.5)	<.001*	−29.2(11.9)	−19.4(15.7)	0.013*
Lateral calcaneal shift T2 [% calcaneal width]	−29.6(10.0)	−16.8(8.2)	<.001*	−32.4(11.3)	−19.4(14.5)	0.007*
Forefoot abduction T1 [°]	−2.4(5.8)	8.9(7.2)	<.001*	−1.3(5.0)	2.7(5.2)	<.001*
Forefoot abduction T2 [°]	−3.6(6.5)	10.2(8.0)	<.001*	−3.9(4.0)	2.1(5.7)	<.001*
Arch height T1 [% foot length]	9.8(3.5)	14.1(5.1)	<.001*	9.1(2.9)	10.3(3.0)	0.193
Arch height T2 [%foot length]	12.4(3.8)	16.9(4.3)	<.001*	11.5(3.4)	12.6(2.6)	0.227

KAM1: first peak of knee adduction moment. – KAM2: second peak of knee adduction moment. – T1: mean value of parameter at time point of KAM1. – T2: mean value of parameter at time point of KAM2. FFpre: Subgroup of children with flatfoot deformity before surgical correction. – FFpost: after surgical correction.

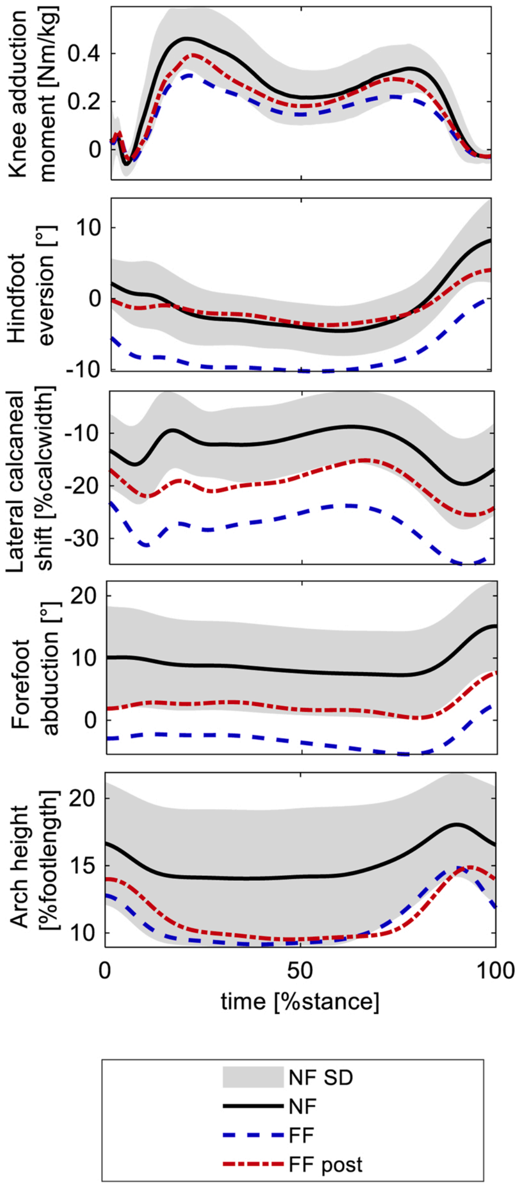

Fig. 2. The knee adduction moment and foot parameters (hindfoot eversion, lateral calcaneal shift, forefoot abduction and arch height) in stance for flat-footed children (FF, dashed blue), for normal footed children (NF, black solid with standard deviation in grey) and flatfooted children after surgery (FF post, dash-dotted red). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

corrective surgery and were re-analysed on average 15 months after surgery. Hindfoot eversion, lateral calcaneal shift and forefoot abduction in stance improved after surgery (Table 2 and Fig. 2) and was accompanied by a significant increase in KAM1 and KAM2 post operatively ($p < .05$). While lateral calcaneal shift was significantly correlated with both peaks in KAM prior to surgery, change in lateral calcaneal shift following surgery was significantly correlated with the change in KAM1 ($r = 0.54$, $p = .02$) but not KAM2 ($r=-0.24$, $p = 0.32$).

4. Discussion

This study investigated the impact of flatfoot deformity on KAM in children during barefoot walking and whether components of flatfoot deformity, namely hindfoot eversion, lateral calcaneal shift were related to KAM1 and forefoot abduction and MLA height to KAM2. Consistent with previous research [3], KAM1 and KAM2 were significantly lower in children with flatfeet than in children with rectus feet. Contrary to our hypothesis, however, neither hindfoot eversion nor forefoot abduction was correlated with KAM1 or KAM2. Rather, lateral calcaneal shift was found to be moderately correlated with KAM1 and tended to be moderately correlated with KAM2 in FF and NF children. MLA height was moderately correlated with KAM2 in FF and NF. While corrective surgery for flatfoot tended to normalize between-group differences in KAM1 and KAM2, changes in lateral calcaneal shift with surgery were found to correlate with only changes in KAM1. Thus, lateral calcaneal shift would appear to be a prime determinant of KAM1 in children during walking and to a lesser extent KAM2, which is also influenced by MLA height.

Altered knee adduction moments during walking in adults have been linked with the development of knee joint pain and disease [10,20]. In this study, FF showed lower internal knee valgus moments during walking than NF children. Both KAM1 and KAM2 were lower in FF, which reflect a relatively greater load bourn by the lateral compartment of the knee. These loading characteristics are similar to those seen in patients with valgus knee alignment [14] and in some studies have been identified as a risk factor for lateral induced osteoarthritis in adults [9,21]. It should be noted, however, that we found no literature to date, implicating flatfoot deformity with lateral knee osteoarthritis. On the contrary, flat feet have been linked with cartilage damage involving the medial knee compartment [22]. Further, Levinger et al. reported a greater occurrence of a “pronated foot posture” in patients with medial knee osteoarthritis based on static measurements of foot shape [23]. In a later study, these authors also showed that peak hindfoot eversion was significantly correlated with KAM2 in elderly patients with medial knee osteoarthritis [24]. While we observed no significant relationship of KAM to hindfoot eversion, it is important to note that we evaluated the relationship of hindfoot eversion to KAM at the time of peak KAM occurrence rather than comparing peak values.

The results of the current study also suggest that a lateral shift of the calcaneus results in lower KAM1, whereas both, lateral calcaneal shift and a low MLA height is associated with lower KAM2. Previous research has shown that the knee adduction angle and the magnitude of vertical ground reaction force were predictors of KAM1, rather than the location of initial heel contact [25]. In the current study, FF and NF children had similar vertical ground reaction forces and knee adduction angles, as verified by inclusion of only children with normal hip and knee alignment. Therefore, our results suggest that the point of force application due to the calcaneal shift, rather than the calcaneal tilt per se, may play an important role in lowering peak KAM. In support of this concept, shoe modifications that produce a lateral shift in the point of ground reaction have been shown to reduce KAM, particularly KAM1, in healthy young adults [26].

Contrary to our hypothesis, forefoot abduction was not related to KAM2 rather both lateral calcaneal shift and low MLA height appeared to contribute to the lowering of KAM2. The finding that arch height was related to KAM2 in FF is consistent with previous findings in the literature [3]. Although the precise mechanism behind the link remains obscure, one possible explanation could be related to a medial thrust of the knee during the push-off phase of gait, which could result from a medially rotated tibia as seen in this cohort. Further exploration of our data showed an inverse correlation of knee rotation and KAM2. This finding is in agreement with the literature showing a reduction of KAM2 in healthy adults walking with a “medial thrust” gait modification strategy [27]. On the other hand, KAM2 improved after flatfoot correction while arch height did not significantly increase.

In terms of practical research, KAM is an important parameter as it is an indicator of knee joint loading and has been extensively investigated in the field of clinical gait analysis [10–14,20]. Several gait modifications and orthopedic malalignments, such as trunk lean and in-toeing, are known to influence KAM [28]. The difference in KAM1 and KAM2 in children with flat rather than rectus feet in this study (≈30 %) is similar to that previously induced with a medial knee thrust gait pattern

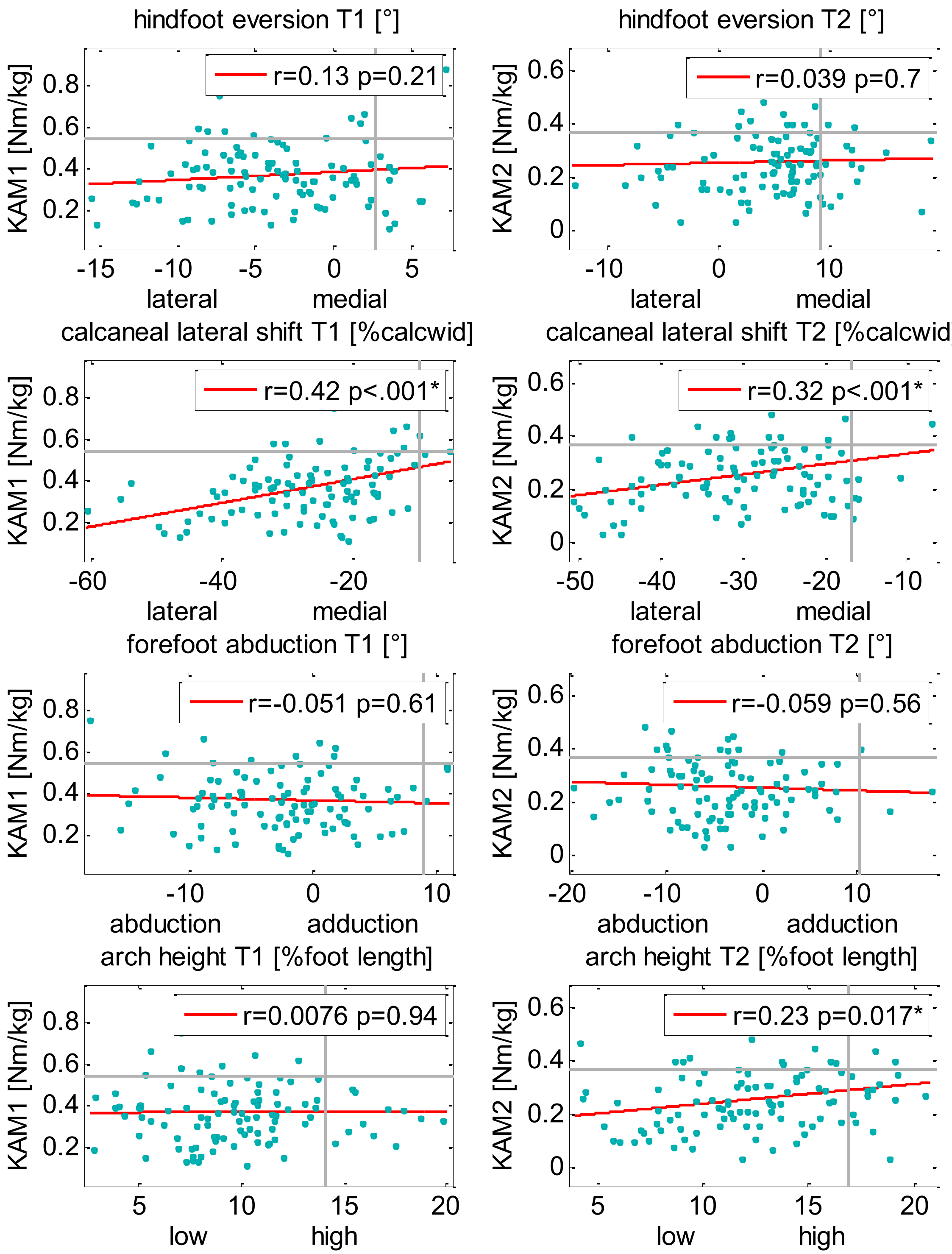

Fig. 3. Linear relationship of flatfoot parameters and KAM peaks with mean of norm relationship (grey cross) with corresponding correlation coefficients (r) and p-values. Significant results are marked with *.
KAM1: first peak of knee adduction moment. – KAM2: second peak of knee adduction moment. – T1: parameter value at the time point of KAM1. – T2: parameter value at the time point of KAM2. – %calcwid: percentage of calcaneal width.

(20 %) [11], an excessive trunk lean (20 %) [29], valgus knee alignment (30 % KAM1) [14], and a toe-in (45 % KAM1) or toe-out (56 % KAM2) gait pattern [13]. Hence, flatfoot deformity might have a clinically significant impact on KAM and knee joint loading. The results of this study also highlight that calcaneal shift, rather than more conventional parameters of foot alignment, should be considered when evaluating KAM. As evidenced in this study, none of the standard parameters showed any relationship with knee adduction moments at time point of peak KAM occurrence.

In the current study, step width was significantly greater in FF children rather than NF children. Step width is known to affect KAM. Intentional increases in step width in healthy persons has been associated with lower KAM [30]. The lower KAM observed in FF, therefore, could reflect their greater step width (2.9 ± 0.7 cm) compared to those with NF during walking. However, step width remained unchanged in the 19 children that underwent corrective surgery despite an improvement in KAM1 and KAM2 following surgery. Hence, at least in this study, step width would appear to have had negligible influence on KAM in these children.

Considering further limitations, the retrospective nature of this study likely introduced significant selection bias, as children with flatfoot presenting for gait assessment at a children's hospital may not be representative of the wider community and those receiving surgical correction of flatfoot may present with more severe deformity or when mild than with severe pain. Moreover, post-surgical functionality was not specifically assessed in this study; although following surgery children were able to walk at the same, if not faster, speeds than prior to surgery. Further, KAM is influenced by gait speed [28]. While children in this study were free to adopt their preferred walking speed, the results may not be relevant at higher or lower speeds. Additionally, as with all optical motion analysis methods, the calculation of calcaneal shift is based on skin mounted markers and may be subject to error. Finally, there might exist a potential inter-relationship between the measurement of OFM hindfoot eversion, which assesses the hindfoot motion relative to the tibia, and the measurement of lateral calcaneal shift, which describes the lateral shift of the hindfoot between the ankle joint and calcaneus center. Both hindfoot valgus and lateral calcaneal shift may coexist in a flatfoot and may be clinically difficult to distinguish. The introduction of the new parameter, lateral calcaneal shift, in this study is an attempt to discern one deformity from the other. The lateral calcaneal shift is taken in addition to standard clinical findings in the surgical decision making process of a symptomatic flatfoot deformity. However, further research is necessary to evaluate the reliability of this parameter.

5. Conclusion

In conclusion, this study found that children with flatfoot have lower KAM peaks than children with a rectus foot. While KAM was unrelated to traditional gait parameters such as hindfoot eversion and forefoot abduction, MLA height and a relatively new measure, lateral calcaneal shift, contribute to a lower KAM in children during walking. Lateral calcaneal shift and medial longitudinal arch height may, therefore, be useful measures for monitoring the effects of foot posture on KAM in children during walking. Hence, the calcaneal shift should be considered as a standard measure in clinical gait analysis.

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CRediT authorship contribution statement

S. Kimberly Byrnes: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Project administration, Software, Validation, Visualization, Writing - original draft. **Scott Wearing:** Methodology, Supervision, Visualization, Writing - review & editing. **Harald Böhm:** Conceptualization, Data curation, Funding acquisition, Investigation, Methodology, Software, Supervision, Visualization, Writing - review & editing. **Chakravarthy U. Dussa:** Funding acquisition, Investigation, Supervision, Visualization, Writing - review & editing. **Thomas Horstmann:** Methodology, Supervision, Visualization, Writing - review & editing.

Declaration of Competing Interest

All authors do not have any financial or personal relationships with other people or organizations that inappropriately influence this work.

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