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The Lower Extremity Kinematics and Kinetics of Stationary Cycling in Young Children With and Without Cerebral Palsy

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The Lower Extremity Kinematics and Kinetics of Stationary Cycling in Young Children With and Without Cerebral Palsy

by

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Dissertation

Presented to the Faculty of the Graduate School of

The University of Texas at Austin

in Partial Fulfillment

of the Requirements

for the Degree of

Doctor of Philosophy

The University of Texas at Austin

May, 2016

The Lower Extremity Kinematics and Kinetics of Stationary Cycling in Young Children With and Without Cerebral Palsy

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The University of Texas at Austin, 2016

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Children with cerebral palsy (CP) are at risk of secondary changes, such as bone deformation. Physical activity is important to reduce these changes and to improve physical function, especially when children are young. Cycling is a great locomotor skill for this because it can be used in therapeutic and in recreational settings. Standardization of cycling protocols is needed to give therapists a better understanding how maximum improvements can be obtained. For this, we have to understand the biomechanics of cycling in young children with CP.

We have investigated the kinematics and kinetics of cycling in young children with and without CP, and the influence of changes in task demands on these biomechanics. It was hypothesized that young typically developing (TD) children have altered biomechanics during cycling in comparison to older children and that young

children with CP have alterations in comparison to TD peers. Furthermore, it was expected that spasticity reduction by botulinum toxin (BTX) treatment would improve these biomechanics in children with CP.

In Study 1, it was shown that 4-year-olds TD children had more out-of-plane motion than 6-, 8- and 10-year olds, while motions in the sagittal plane were similar between groups. Also, 4-year-olds produced higher jerk cost when cycling at high cadences. In Study 2, analysis revealed that children with CP were only able to cycle at low resistance and low cadences and were less adaptable to changes in movement speed. Adjustments in joint angles, joint torques, jerk cost and pedal forces in all three planes of motion were observed, indicating that children with CP cycled less efficiently than TD peers. In Study 3, it was shown that CP-specific alterations in cycling biomechanics were reduced 3 weeks after BTX injections.

This work has increased our understanding of the cycling ability of young children. It led to the conclusion that children with CP can cycle under reduced task demands in comparison to TD peers. When spasticity is reduced, the adaptability of children with CP against changes in task demands is increased. The findings of these studies help us understanding the influences of cycling in children with CP.

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Chapter 1: General Introduction

CEREBRAL PALSY

In the study of motor development, investigators have been interested in how motor behavior changes during human development and the processes that lead to these changes (Clark & Whitall, 1989). Different disorders, such as cerebral palsy (CP), Down syndrome and developmental coordination disorder, can influence motor development from an early age (Hadders-Algra, 2010; Vicari, 2006). CP, for example, can lead to many physiological symptoms, such as cognitive and motor impairments (Paneth, 2008), often resulting in children with CP reaching defined motor development milestones later than typically developing (TD) children, if they reach them at all. CP can affect a broad range of motor skills such as independent walking and bimanual fine motor activity, and it can also reduce strength (Damiano, 2009; Dodd, Taylor, & Damiano, 2002; Facchin et al., 2009).

CP is a non-progressive disorder as the brain insult does not increase in severity over time (Paneth, 2008). However, primary effects (e.g. muscle spasticity and imbalance between muscle agonists and antagonists) and secondary effects (e.g. bone deformation and reduced muscle growth) can be progressive (Gage & Novacheck, 2001). Early therapeutic intervention and regular participation in physical activity are necessary to improve the motor development of children with CP to delay surgery and to reduce the incidence of secondary effects (Damiano, 2006; Graham et al., 2000; Landsman, 2006).

In addition to the reduction of secondary risk factors, an active lifestyle promotes the development of fundamental motor skills typical of childhood (Anderson, Spencer-Smith, & Wood, 2011) and functional skills necessary for physical independence.

The independent upright posture becomes more challenging, for children with CP as they grow and the manifestation of the neuromotor deficits changes. Walking becomes more challenging and the child becomes more dependent upon mobility devices. The tricycle and bicycle employ the same repetitive cyclic motion of the lower extremities and enable children with CP, to engage in activities that give them more opportunities to be physically active and socially engaged.

In the therapeutic environment, cycling can offer an early opportunity for intensive intervention as children with CP may be able to translate the therapeutic activity to a recreational environment (Fowler et al., 2007). Many young children with CP have the ability to pedal a tricycle or bicycle, even if they are unable to walk because cycling is less dependent on balance than walking (Fowler et al., 2007). Riding a tricycle requires only maintaining upper body balance and the arms provide an additional point of contact for stability. Walking, on the other hand, does not provide for hand support and the child has to manage lower extremity control while aligning the trunk over the moving base of the hips. When needed, trunk and head support can be provided on a tricycle, to reduce the demand for balance further (eSpecialNeeds, 2015).

Cycling has been shown to contribute to increased muscle strength and improved functional ability, such as standing and walking, in children (between 6-12 years) and adolescents with CP (Chen et al., 2012; Williams & Pountney, 2007). Despite the physical training benefits associated with cycling, there is no standardized regimen for cycling therapy for children with CP. Cycling in a therapeutic setting varies from cycling for recreation. Cycling for therapy is not a self-paced activity, and it is prescribed in terms of pace, resistance, and duration. It is unclear from the literature whether cycling is being used for muscle strengthening, aerobic capacity training, or technical training. Independent of the goal, the repetitive nature of cycling and reduced periods of rest can increase existing problems, such as pain, fatigue and hip and knee injuries. Standardization of cycling protocols is needed to optimize the benefits gained for the goal in mind.

Motor impairment due to CP is expressed as an alteration in performance biomechanics. Cycling is typically thought of as a planar (sagittal plane) motion. Studies have shown, however, that older children and adolescents with spastic CP usually display more movement out of the sagittal plane when compared to age-matched peers (T. E. Johnston, Barr, & Lee, 2007; T. E. Johnston, Prosser, & Lee, 2008). To date, the kinematics and kinetics of cycling have not been studied in *young* children with CP, yet such research is especially important because early therapeutic intervention is needed for a successful habilitation of children with CP. From studying TD children, we have learned that the biomechanics of older children cannot be generalized to younger children. Both, young and old, typically- and atypically-developing children can make the pedal go around, but how they achieve that task differs. To understand atypical patterns of movement production, we must first understand the changes in production that occur in early typical development.

Cycling biomechanics during typical development

It is widely appreciated that as a child grows and matures there are significant changes to the biomechanics of cycling. Younger children (< 7 years) are more variable with cadence and consistency of speed in moving the crank around the cycle (Jensen & Korff, 2004). When cadence increases, these younger cyclists alter the power production strategies and break the typical of hip to ankle synergy and begin to generate more power through the knee musculature (Korff & Jensen, 2007). Changes in kinematics, such as out-of-plane motion and reduced movement fluency, are not fully understand in TD cycling so far while these are attributed to the atypical development of children with CP. Considering that older children and adolescents with CP both show alterations in these parameters during exercise of locomotor skills (T. E. Johnston et al., 2007; Kaplan, 1995; Wren, Rethlefsen, & Kay, 2005), it can be assumed that the altered motion is primarily due to the effects of CP.

However, before studying the changes in kinematics in young children with CP, it is important to investigate if changes in kinematics are due to the atypical development of children with CP or whether they are features of typical development due to the immature neuromuscular system. Concerning gait, Sutherland et al. showed that 1-year-old TD children who are learning to walk, have more out-of-plane motion and more external hip rotation and hip abduction than children 2 years or older (Sutherland, Olshen, Cooper, & Woo, 1980). Because no other studies have investigated this phenomenon in connection with any other locomotor skill, the purpose of the first study was to determine whether young TD children have different kinematics during cycling in comparison to older TD children. With the investigation into the motor development of young TD children during cycling, it was then possible to determine to what extent the altered kinematics in young children with CP are typical of novice cycling and an unpracticed motor system for this task, or a function of the morphological and neurological impact of cerebral palsy. This information can be used to guide therapeutic strategies – are we working with an immature system in building a strong referent of correct performance or attempting to overcome and alter the performance to accommodate structural and functional changes induced by cerebral palsy?

BIOMECHANICS DURING CYCLING IN CHILDREN WITH CEREBRAL PALSY

With knowledge about the development of TD children in cycling, we were able to investigate how children with CP cycle differently than TD children. From cycling studies performed with older children (around 10 years of age) and adolescents with CP, it is known that these populations produce more out-of-plane motion (T. E. Johnston et al., 2007), they show lesser fluency in motion (Kaplan, 1995), more co-contraction (T. E. Johnston et al., 2007; Kaplan, 1995) and apply less efficient forces to the pedals (T. E. Johnston et al., 2008). Considering that young TD children cycle differently than older TD children and that children with CP have altered motor skills because they decline in ambulatory ability as they get older (Bottos & Gericke, 2003), the cycling biomechanics of young children should be investigated for early intervention. This is of particular importance from a therapeutic viewpoint because the efficacy of cycling therapy requires that it be optimized to address the demands of each age group.

The literature provides a clear picture that those with CP vary in the performance of cycling compared to their TD peers. Importantly, we must understand how cycling is constructed – what are the muscular forces (torques in the case of segmental rotations) that contribute to moving the crank.

Therefore, in the second study, the kinematics and kinetics during cycling in *young* children with CP were analyzed. This study highlighted the biomechanical changes in children with CP, and its findings provide clinicians with a better understanding of the effects of cycling on the lower extremities.

EFFECT OF BOTULINUM TOXIN ON CYCLING BIOMECHANICS

Functional performance is hampered in children with CP because of spasticity. Spasticity is defined as an abnormally increased level of muscle activity (hypertonia) during rest and an increased resistance during passive movement (Sanger et al., 2007). The high resistance is caused by hyperactive stretch reflex mechanisms (Rethlefsen, Ryan, & Kay, 2010). Increased levels of muscle activity lead to altered coordination during fine and gross motor skills.

Alterations in the biomechanics of young children with CP lead to inefficient cycling and potentially damaging secondary effects. The question remains how therapies that reduce spasticity alter the cycling performances of children with CP? Spasticity reduction would allow for the use of more effective movement strategies during cycling. Increased adaptability means that children with CP would be able to cycle comfortably at a variety of cadences.

Spasticity is susceptible to various treatments. For example, physical and occupational therapy can reduce the severity of muscle tone and improve patients' ability to move their limbs (M. V. Johnston, 2009; Tilton, 2009). Also, other methods, such as botulinum toxin (BTX) injections, oral medication, selective dorsal rhizotomy and orthopedic surgery, have been shown to extend the improvements made by physical and occupational therapy (Grotta et al., 2004; Msall & Park, 2008; Tilton, 2009). BTX injections are a commonly used treatment for spasticity in young children with CP. The pharmacological activity of BTX is based upon the capability of the toxin to block nerves from releasing acetylcholine to the muscle, preventing the spastic muscle from contracting (Arnon et al., 2001; Koman, Smith, & Shilt, 2004). BTX is a desirable treatment because it can target specific muscles that are most affected by spasticity, and the effect of BTX is reversible, with effects lasting 2 to 4 months (Koman et al., 2004; Love et al., 2010).

We would expect to see many improvements in the efficiency of cycling after BTX treatment because of its effectiveness in reducing spasticity. While there have been no formal studies of the impact of BTX on cycling, it has been shown to reduce joint torques in the ankle during gait and improve movement fluency during reaching (Bensmail, Robertson, Fermanian, & Roby-Brami, 2010; Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000). The question remains whether BTX can also lead to improved biomechanics in other motor skills, such as cycling, with emphasis on joint stress and movement fluency. In Study 3, the effects of BTX on the kinematics and kinetics of young children with CP were studied during cycling at different cadences.

OUTLINE OF THE DISSERTATION

The purpose of the three studies presented in this dissertation was to examine the developmental biomechanics of young children with and without CP during sub-maximal cycling and the influence of BTX injections on these characteristics. In Study 1, we sought to identify age-related changes in the three-dimensional biomechanical characteristics of cycling as performed by TD children (Chapter 2). This study provides the early developmental comparison case for evaluating the cycling ability of children with CP. In Study 2, we observed the consequences of neurological insult and spasticity on the three-dimensional biomechanics of cycling in young children with CP as compared to their age-matched TD peers (Chapter 3). At last, in Study 3, we assessed the benefits obtained from treatment with BTX. BTX has been shown to be effective in reducing muscle spasticity. If you take the spasticity away, how does the child move the crank differently? It is this information that will help the therapist understand more clearly the benefits of cycling therapy under the influence of BTX as adjuvant treatment for functional movement training (Chapter 4).

Chapter 2: Study 1

The development of cycling coordination in young typically developing children

INTRODUCTION

Cycling is a common form of recreation during childhood and is now emerging as a therapeutic intervention to habilitate those with developmental delay and rehabilitate those with acute injury or illness. There are many advantages to using cycling for rehabilitation. Seat support reduces balance demands, and cycling provides a viable means of exercise and enables independence in locomotion, which impacts both the physical and mental well-being of an individual (Fowler et al., 2007; Jansen, van Alfen, Geurts, & de Groot, 2013; Johnston, Barr, & Lee, 2007; Mutton et al., 1997; Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). For children and adolescents with cerebral palsy (CP), cycling intervention is becoming commonplace as a means for improving functional skills such as independent standing and walking (Johnston, Prosser, & Lee, 2008b; Kaplan, 1995).

Past research has shown that children and adolescents with CP are inefficient in their efforts to move the crank through a complete 360^{o} revolution. Adolescents with CP cycle less fluently, have atypical kinematics and are less efficient in delivering power to the crank (Johnston et al., 2007; Kaplan, 1995). Interestingly, these are the same attributes that are used to describe young children who are beginning to learn how to pedal a bicycle. From a rehabilitation perspective, we look at these inefficiencies and extraneous movements as targets for therapeutic intervention. From a developmental perspective, such inefficiencies and extraneous movements are typical of inexperience and/or an immature biological system. When the developmentally immature child is also afflicted with CP what markers distinguish typical development from the manifestations of disease? What is the appropriate target for clinical intervention? The purpose of this study was to describe the emergence of cycling skills in typically developing (TD) children, to provide a baseline against which similar skills in young children experiencing cycling as a therapeutic intervention can be compared.

What is known about the biomechanics of "typical" cycling emerged first in the studies of elite cyclists, research intended to improve performance and eliminate stress injuries (Daly & Cavanagh, 1976; Hull & Jorge, 1985). A better understanding of the biomechanics of elite cycling provided a standard against which emerging cycling skills could be evaluated, and researchers began to focus on the emergence of cycling skills during typical childhood. Riding a bicycle is a skill that children begin to practice early in life, a cultural phenomenon aided by bicycle developers building tricycles for children as young as 9 months of age. In one of the first studies of its kind, Myrtle McGraw studied the motor development of the twin boys Johnny and Jimmy (McGraw, 1935). McGraw allowed Johnny to practice riding a tricycle daily when he was 11 months of age. At this age, Johnny had no idea how to move his feet to make the pedal go around. After a couple of months, Johnny began pushing on the pedals, but instead of cycling, he started with pushing down with two feet simultaneously, and it was not until 21 months of age that he was able to cycle by alternately pushing his feet downwards.

While Johnny started cycling before he turned 2 years of age, most children are only beginning to cycle at 3 or 4 years of age (Mozer, n.d.). As with most motor skills, early attempts at cycling by children younger than 7 years of age are inefficient and of limited range. Young children (5 to 7 years) are less efficient in transferring hip powers to the crank during the down stroke at high cadences than older children (8 to 10 years) (Korff & Jensen, 2007). To compensate for this lack of efficiency, children deliver more energy directly to the crank during the upstroke by using their knee flexors. Young children also show more variation in the movement characteristics of cycling with larger variability in joint velocities (Jensen & Korff, 2004), an inability to maintain a constant cadence when compared to older children and adults (Liu & Jensen, 2012) and a lower maximum cycling speed compared to adults (Chao, Rabago, Korff, & Jensen, 2002).

What we know about the cycling skills of children is largely limited to the analysis of movement in one plane. Cycling is generally considered a planar motion in the sagittal plane (Hull & Jorge, 1985; Raasch, Zajac, Ma, & Levine, 1997). Typically, the cyclist is seated, and there is a constant contact of the feet with the pedals. In this configuration, the foot/pedal system moves about a circular trajectory while the legs appear to pump up and down. Cyclists will, however, deviate from planar motion under different circumstances. For example, when a cyclist is fatigued, increased out-of-plane motion emerges (Dingwell, Joubert, Diefenthaeler, & Trinity, 2008). Spasticity is also a causative factor of out-of-plane motion in cases of atypical development in children and adolescents (Johnston et al., 2007). In adults, the out-of-plane motion is typically temporary as when fatigued, the out-of-plane motion shown by young children is more persistent as bipedal locomotor abilities are newly emerging skills. In gait, for example, the walking patterns of children in their first-year show more out-of-plane motion than the patterns of older children (Sutherland, Olshen, Cooper, & Woo, 1980). Thus, the out-of-plane motion is one of the parameters that can indicate immaturity in motor skills as seen in gait and reaching (Konczak & Dichgans, 1997; Sutherland et al., 1980).

Cycling a bicycle is a constrained task regarding space – the foot is fixed to the pedal, the pedal to the crank, and the crank turns about a center spindle. Variations in movement characteristics, however, creep into the task as a function of out-of-plane motion and changes in the angular velocity of the crank. The changes in angular velocity may be functional, as in accelerating the crank during the power production phase. Alternatively, such changes may result from poor control of cycling speed. Variability in angular velocity, which is increased in young children (Liu & Jensen, 2012), is an indicator that the jerk cost of a cycling motion will also be increased for this young cycling population. The calculation of jerk cost quantifies these fluctuating cycling velocity characteristics. Jerk cost is also correlated with physiological demand (Fujii & Nagasaki, 1995). Using jerk cost, then, we can document changes in movement efficiency across the age of the cyclist.

In this study, we will compare the kinematics of cycling between younger and older children. We expand upon the existing literature by focusing on the out-of-plane motion and jerk cost associated with non-fatigue cycling. Cadence and resistance are significant modifiers of cycling performance, and these variables can exacerbate biomechanical differences between groups. In this study, we manipulate movement speed to analyze the influence of cadence on the skill construction during early skill acquisition. These data reveal the development of a mechanical solution to a locomotor task in TD children.

METHODS

Participants

Thirty-two TD children participated in this study and were divided into four age groups: 4, 6, 8 and 10 years of age. Children were excluded from the experiment if they suffered from any childhood disease that could influence the motor development or if they had suffered from lower limb injuries within the last 6 months. After explaining the experimental procedures, the parents and children older than 7 years of age signed an informed consent approved by The University of Texas Institutional Review Board.

Four children, one in the 6-year-old group, one in the 8-year-old group and two in the 10-year-old group were excluded from the data analysis because there was too much noise in the marker data to determine the marker positions. Table 2.1 shows the group averages of all the children who were included in the final analysis.

Data preparation and collection

The parents completed a questionnaire about the participants' cycling history. This survey was used to estimate the number of hours the participant cycled in the previous 5 years (Jensen & Korff, 2004). The participants completed the Bruininks-Oseretsky Test of Motor Proficiency, 2nd edition (BOT-2) Short Form at the beginning of the session.

Anthropometric measurements were obtained from each child (height and weight). Furthermore, leg length, circumferences, and skin folds were obtained to predict instantaneous peak power by using a lean thigh volume method established by Martin et al. (Martin, Farrar, Wagner, & Spirduso, 2000). Participants performed the cycling trials at 10% of predicted maximal power (Table 2.1).

The distance between the left and right crank is a fixed distance on the ergometer. To ensure that crank width did not lead to increased movement in the younger children's legs, we calculated the angle between a vertical line running through the anterior superior iliac spine (ASIS) to the ground and a line connecting the ASIS and the center of the pedal, where the foot was clipped in (Figure 2.1). This leg angle was calculated when the foot was at top dead center (TDC).

In order to prepare the participants for data collection, 16 reflective markers were affixed to each participant's body and the pedals: right and left head of the second metatarsal, right and left calcaneus, right and left lateral malleolus, upper third of the right tibia, lower third of the left tibia, left and right lateral femoral condyle, upper third of the right thigh, lower third of the left thigh, right and left greater trochanter, right and left ASIS. One marker was also attached to the pedal spindle on each side. The markers were 14 mm in diameter, and their coordinates were recorded by a 10-camera passive marker tracking system (Vicon Nexus 1.5.1).

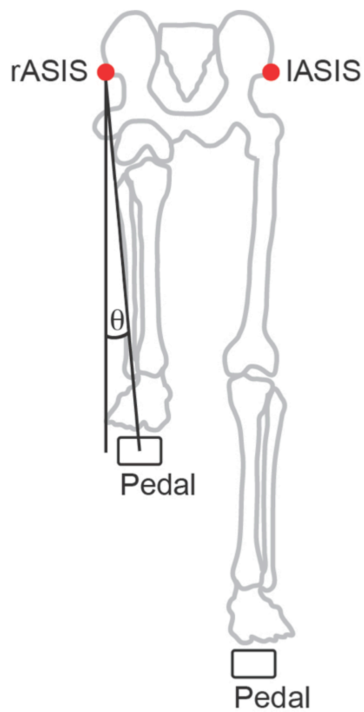


Figure 2.1: Explanation for the calculation of the leg angle. The leg angle is measured between the vertical line from the right ASIS to the floor and the line between the rASIS and the center of the pedal when the right foot was at TDC.

After preparation, the participants were seated upon a stationary cycle ergometer (Lode Corival Pediatric, Lode, Groningen, Netherlands). Position on the ergometer was standardized. Each participant was fixed to the bike using clipless pedals. The crank length was adjusted to 20% of the leg length of the participant. Leg length was defined as the distance from the right greater trochanter to the ground. When participants were seated on the bike and the right foot was positioned at TDC, the seat height was adjusted so that the right knee angle was 75° ±3° and the left knee angle was 155° ± 3°. The handlebars were altered such that the trunk angle was 60° ±3° from the horizontal plane.

The participants were given a target cadence, and they were instructed to stay on target within a tolerance of ±4 RPM. In previous research, it was found that this range is sufficient to identify age-related performance levels (Liu & Jensen, 2009). The participants practiced the cycling tasks without resistance for a total of 5 minutes (cadence feedback was shown on a display on the ergometer). After this practice, which simultaneously served as a warm-up, the participants were asked to cycle at 40, 60, 80, 100 and 120 RPM at a resistance of 10% of their predicted maximum power. The order of the cadences was randomized using a randomization function in Matlab (7.0.1). Participants were asked to cycle at the target cadence until sufficient data were collected (25 seconds). Experimental marker data were collected at a frequency of 120 Hz. At least 1 minute of rest was given between the trials, to avoid fatigue.

Table 2.1: Group characteristics of participants included in the data analysis.

Age group	n (F/M)	Age	Height (m)	Mass (kg)	10% Predicted maximal power (W)	Leg angle (degrees)	Cycling hours	BOT scores (%)
4	8 (5/3)	4yr 7m (5m)	1.07 (0.06)	18.99 (1.85)	8.3 (2.6)	10.8 (2.4)	166 (207)	83 (26)
6	7 (3/4)	6yr 8m (3m)	1.22 (0.06)	23.07 (2.54)	14.3 (6.0)	8.1 (1.2)	291 (254)	77 (14)
8	8 (2/6)	8yr 6m (5m)	1.37 (0.06)	31.11 (5.44)	21.9 (10.7)	6.8 (1.8)	341 (471)	82 (13)
10	5 (2/3)	10yr 7m (3m)	1.46 (0.07)	34.13 (3.58)	27.2 (9.0)	5.7 (2.7)	325 (435)	65 (24)

Note: Means (±SD) are displayed.

Derivation of dependent variables

The data collected were processed using Matlab (7.0.1) and OpenSim (3.0). The kinematic data were low-pass filtered with a cutoff frequency of 20 Hz in Matlab. The right pedal marker was used to determine the cadence of every revolution; each revolution was defined as the motion from TDC until the foot reached TDC again. Five consecutive revolutions within ±4 RPM of the target cadence were used for further analysis, and a trial was omitted if this was not achieved.

Joint angles in three dimensions for each trial were determined using OpenSim (Delp et al., 2007). The musculoskeletal model gait2392_Simbody (Delp et al., 1990) was used with a few adjustments:

1. The pelvic bone was fixed with respect to the ground, to simulate the position of the pelvis on the seat.
2. The knee was remodeled to enable adduction/abduction motion and internal/external rotation in the knee (Donnelly, Lloyd, Elliott, & Reinbolt, 2012).
3. For each participant, the musculoskeletal model of the leg segments was scaled to the length of the lower extremity segments of the participants, based on the marker data that was collected during the experimental session.

The inverse kinematics tool in OpenSim was used to estimate hip and knee flexion/extension, ad/abduction, and internal/external rotation, ankle dorsi/plantar flexion and subtalar pronation/supination angles using the experimental marker data. The hip internal rotation was defined as the inner rotation of the femur about the pelvis and hip abduction was defined as the femur moving away from the midline in the frontal plane. Internal knee rotation was defined as the inner rotation of the tibia about the femur and knee abduction was defined as the outward motion in the frontal plane of the tibia about the femur. OpenSim uses a least squares approach that minimized the differences between the markers from the experimental data and the markers from the model. Per time frame, the model was set in a position that closely matched to the experimental data. In this position, joint angles were estimated from the joint centers-of-rotation (Delp et al., 2007). The maximum and minimum joint angles and the range of motions were determined for each revolution. Within each trial, outcomes parameters from each revolution were averaged. To display the angular motion per group over the crank cycle, the motion data for each revolution were re-sampled to exhibit the same number of data points per revolution so the revolutions could be averaged per trial and group.

For each revolution, the crank angle was calculated using the right pedal marker. Per revolution, jerk was computed as the third derivative of the crank angle. Jerk cost was calculated using equation 1:

$$Jerk\ Cost = {}^{1}/_{2} \int_{0}^{T} J(t)^2 dt \qquad \text{(Eq. 1)}$$

where J(t) is the jerk per revolution and T is the revolution movement time. For each trial, the jerk cost of five revolutions was averaged.

Statistical analysis

To establish any differences across the four age groups, statistical analyses were performed using SPSS Statistics 21 with significance levels set at $p<0.05$. Previous cycling hours, BOT scores, and leg angle were separately analyzed using one-way ANOVAs, to determine whether any confounding effects were leading to group differences. Linear mixed models with a repeated subject effects were used to determine the maximum and minimum value and the range of motion of each joint angle and the jerk cost calculated from the crank angle motion. Age and cadence were included in the model as fixed factors.

We tested the hypothesis with regard to age-related differences in cycling performances by investigating the main effect of age. Furthermore, we investigated the interaction effect between age and cadence, to study the age effect on cycling speed in joint kinematics. A Bonferonni correction was applied to reduce the chance of Type 1 errors; the significance level was divided by the number of joint angles (for example, $p < 0.05/8 = p < 0.006$). In the case of a finding of significance in the linear mixed model, Tukey HSD post hoc tests were performed to analyze the differences within the two fixed factors and the interaction effect. For these post hoc tests a significance level of $p<0.05$ was used. All successful trials were included in the analysis.

RESULTS

Confounding effects

No participants scored below average on the BOT-2, thus, no participants were excluded from the study based on his or her score. The group averages of the prior cycling experience hours and the BOT-2 scores (Table 2.1) were compared between groups, to show that there were no confounding effects in kinematics due to experience and level of proficiency. No group effects were found for either BOT-2 or cycling experience ($p>0.05$).

A significant effect was found for the leg angle ($P = 0.001$). Follow-up tests showed that 4-year-olds had a significantly larger leg angle (-10.8°) than members of the 8- and 10-year-old groups (6.8° and 5.7°, respectively, Table 2.1). No differences in leg angle were observed between the 4-year-olds and 6-year-olds and between the 6-year-olds and 8- and 10-year-olds.

Successive trials

All children were instructed to cycle at a steady state at five different cadences, but not all children were able to cycle constantly for the minimum of five consecutive revolutions at each target cadence. The 4-year-olds, in particular, found it more challenging to cycle at a steady state as speed increased. Table 2.2 shows the number of participants within each group who met the revolution minimum at each target cadence When the children reached 120 RPM, for the 4-, 6- and 8-year-old groups, at least one child was unable to perform successfully the trial.

Table 2.2: Number of participants who successfully cycled at steady state at a given target cadence.

Group	40 RPM	60 RPM	80 RPM	100 RPM	120 RPM
4 year	7 (88%)	8 (100%)	6 (75%)	5 (63%)	3 (38%)
6 year	6 (86%)	7 (100%)	7 (100%)	7 (100%)	6 (86%)
8 year	8 (100%)	8 (100%)	8 (100%)	8 (100%)	7 (88%)
10 year	5 (100%)	5 (100%)	5 (100%)	5 (100%)	5 (100%)

Note: Between brackets, the percentage of participants in each group who successfully performed the given task is stated.

Joint angles

It is the 4-year-olds who cycled differently than children in the older age groups. While cycling, the hip and foot are in continuous contact with external structures, i.e. the pedals and seat of a bicycle or ergometer. This leads to a closed motor skill condition under which the joint angles are dependent upon each other. When the femur moved out of the plane of motion (hip abduction), the subtalar joint rotated to move the foot inwards. Significant age main effects ($p < 0.006$) were found for the maximum hip abduction and maximum and minimum supination in the subtalar joint (Table 2.3, Figure 2.2-2.4). The hip abduction was most prominent at TDC. Post hoc tests revealed that 4-year-olds produced more abduction at the hip than 6- and 10-year-olds at this point. When the foot reached the bottom dead center (BDC), hip abduction was reduced in all groups, and the differences in abduction between the groups were nonsignificant (Figure 2.2 & 2.5). The 4-year-olds had greater knee external rotation and abduction than the other groups (Figure 2.3); however, these differences were not significantly different from each other. During the entire cycling motion, the 4-year-olds cycled with more supination in the subtalar joint than all the other groups (Figure 2.4 & 2.5), and they had significantly more supination at maximums and minimums than the 6- and 10-year-olds.

The 4-year-olds had more hip internal rotation movement and more subtalar motion than the other groups. The age main effects for the range of motion (ROM) of the hip internal/external rotation and the subtalar supination/pronation motion were significant (Table 2.3). Post hoc tests revealed that 4-year-olds had more rotation in the hip (Figure 2.2 and 2.5) and more motion in the subtalar joint during cycling than the 6-, 8- and 10-year-olds (Figure 2.4 and 2.5).

No significant interaction effect between age and cadence was found after we applied the Bonferroni correction on the multiple Manovas.

Table 2.3: Statistical analysis of the main effect for age and age X cadence interactions.

Motion	**Age X Cadence Interaction**		**Age Main Effect**	
	F	P	F	P
ROM Hip Flexion	F(12,18.86)=1.154	0.378	F(12,23.10)=2.198	0.115
ROM Hip Abduction	F(12,17.62)=0.946	0.528	F(3,20.10)=3,468	0.035
ROM Hip Rotation	F(12,16.86)=0.789	0.791	F(3,22.16)=7.651	**0.001**#
ROM Knee Flexion	F(12,17.25)=1.441	0.238	F(3,23.57)=3.777	0.024
ROM Knee Adduction	F(12,19.38)=0.885	0.575	F(3,22.65)=1.147	0.351
ROM Knee Rotation	F(12,19.70)=1.522	0.197	F(3,24.06)=0.890	0.460
ROM Ankle Flexion	F(12,24.86)=1.329	0.264	F(3,23.69)=0.971	0.423
ROM Subtalar motion	F(12,20.16)=2.616	0.027	F(3,23.68)=7.251	**0.001**#
MAX Hip Flexion	F (12,23.08)=0.643	0.784	F(3,23.05)=0.521	0.672
MIN Hip Abduction	F(12,22.68)=1.652	0.146	F(3,24.26)=3.278	0.038
MAX Hip Rotation	F(12,21.50)=2.356	0.040	F(3,22.81)=4.286	0.015
MAX Knee Flexion	F(12,24.34)=1.552	0.173	F(3,23.78)=0.989	0.415
MAX Knee Abduction	F(12,15.89)=3.443	0.012	F(3,23.72)=4.085	0.018
MAX Knee Rotation	F(12,18.99)=1.179	0.363	F(3,21.69)=2.303	0.105
MAX Ankle Flexion	F(12,23.97)=1.348	0.257	F(3,23.27)=0.342	0.796
MAX Subtalar motion	F(12,18.51)=0.393	0.949	F(3,22.11)=7.091	**0.002**%
MIN Hip Flexion	F(12,20.21)=1.291	0.296	F(3,23.96)=0.375	0.772
MAX Hip Abduction	F(12,19.25)=1.562	0.185	F(3,23.23)=7.066	**0.002**%
MIN Hip Rotation	F(12,19.78)=1.108	0.406	F(3,23.25)=0.929	0.442
MIN Knee Flexion	F(12,13.79)=1.279	0.328	F(3,23.81)=0.855	0.478
MIN Knee Adduction	F(12,21.03)=1.290	0.294	F(3,23.56)=1.196	0.333
MIN Knee Rotation	F(12,17.51)=0.578	0.832	F(3,23.00)=2.002	0.142
MIN Ankle Flexion	F(12,22.67)=1.104	0.403	F(3,22.87)=0.167	0.918
MIN Subtalar motion	F(12,20.39)=1.117	0.399	F(3,21.84)=5.726	**0.005**%

Note: # 4 > 6, 4 > 8, 4 > 10
% 4 > 6, 4 > 1

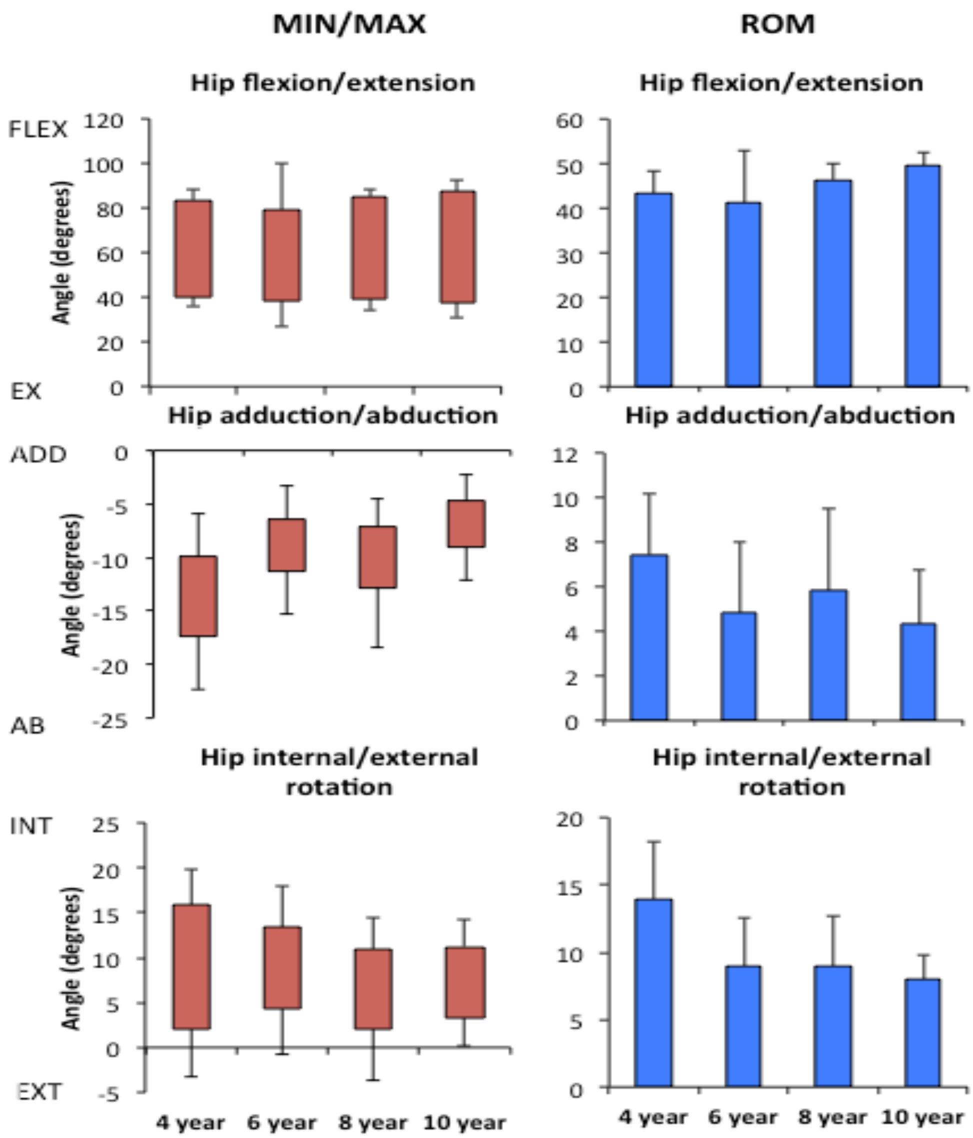


Figure 2.2: Hip angle peaks and range of motions. The bars on the left represent the ROM in the joint angle. The top of each bar represents the maximum rotation and the bottom of the bars the minimum rotation, averaged by group (including all cadences). The error bars on top indicate the standard deviations of the maximum rotation, and the error bars at the bottom indicate the standard deviation of the minimum rotation. The bars on the right represent the ROM in the joint angle. FLEX = flexion, EX = extension, ADD = adduction, AB = abduction, INT = internal rotation, and EXT = external rotation.

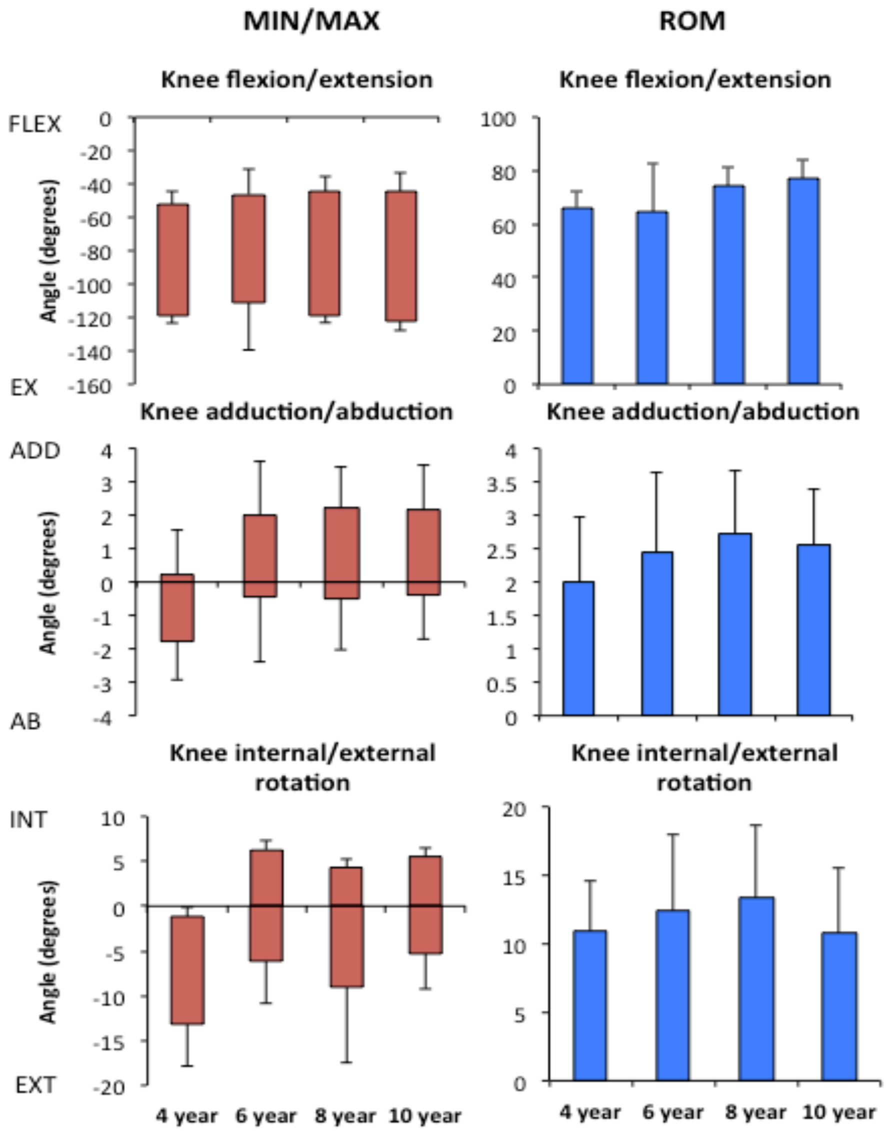


Figure 2.3: Knee angle peaks and range of motions. The bars on the left represent the ROM in the joint angle. The top of each bar represents the maximum rotation and the bottom of the bars the minimum rotation, averaged by group (including all cadences). The error bars on top indicate the standard deviations of the maximum rotation, and the error bars at the bottom indicate the standard deviation of the minimum rotation. The bars on the right represent the ROM in the joint angle. FLEX = flexion, EX = extension, ADD = adduction, AB = abduction, INT = internal rotation, and EXT = external rotation.

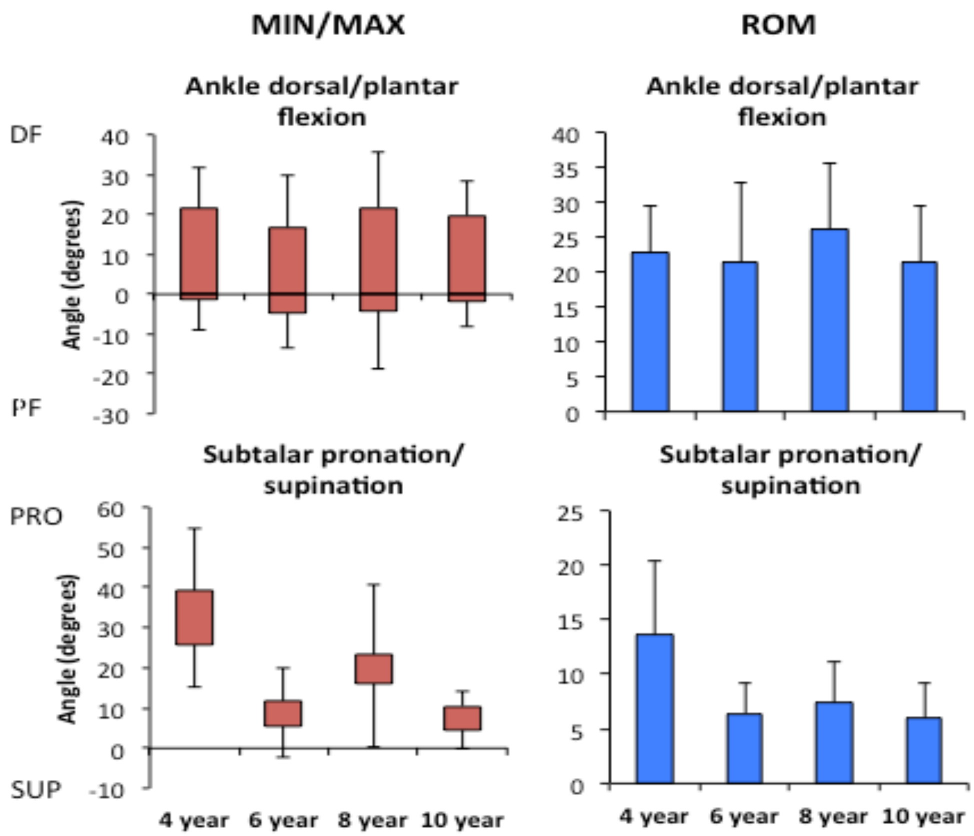


Figure 2.4: Ankle angle peaks and range of motions. The bars on the left represent the ROM in the joint angle. The top of each bar represents the maximum rotation and the bottom of the bars the minimum rotation, averaged by group (including all cadences). The error bars on top indicate the standard deviations of the maximum rotation, and the error bars indicate at the bottom the standard deviation of the minimum rotation. The bars on the right represent the ROM in the joint angle. DF = dorsi flexion, PF = plantar flexion, PRO = pronation, and SUP = supination.

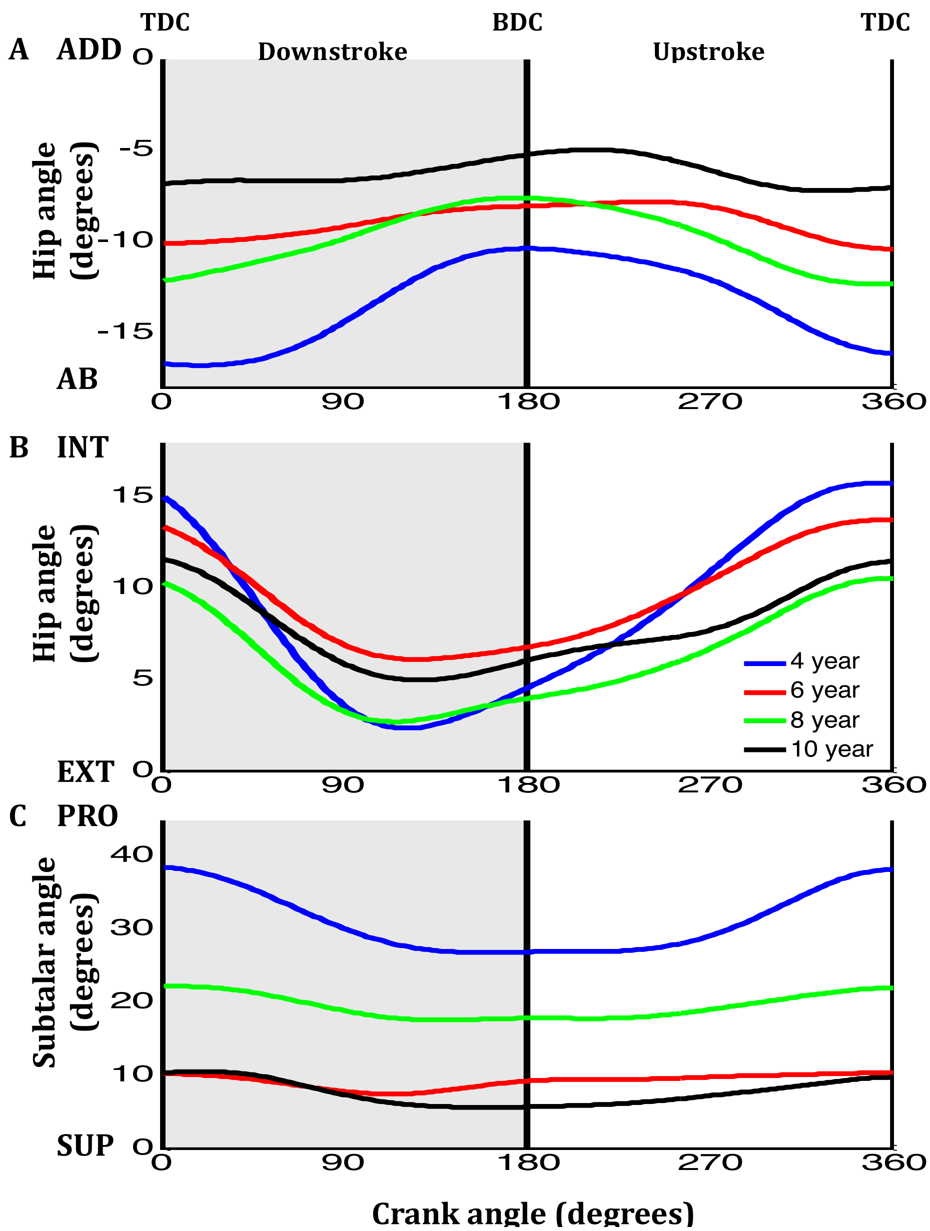


Figure 2.5: Out-of-plane motion. Hip ab/adduction (A), hip rotation (B), and subtalar rotation (C) are shown for each group. The line graphs represent the motion in each particular joint during one crank revolution. Only the joint angles (hip adduction, hip rotation, and subtalar angle) that are significantly different between the children are plotted. ADD = adduction, AB = abduction, INT = internal rotation, EXT = external rotation, PRO = pronation, and SUP = supination.

Jerk Cost

At low cadences, all children produced the same amount of jerk cost. However, when cadence increased to 100 and 120 RPM, the 4-year-olds cycled with less fluently, with more jerk cost, than 6-, 8- and 10-year-olds. A significant age main effect ($p<0.001$) and age X cadence interaction effect ($p<0.001$) were found for jerk cost. The significant interaction effect clearly indicates that children only differed from each other when they cycled at higher cadences. At 100 RPM, the 4-year-olds were producing significantly more jerk cost than the 8- ($p=0.001$) and the 10-year-olds ($p<0.001$). When the children cycled at 120 RPM, the 4-year-olds produced more jerk cost than all three other groups ($p<0.001$ for each comparison). The reduction in fluency of cycling was clearly a function of the extremes in cadence. No differences were found between the groups at 40, 60 and 80 RPM (Figure 2.6).

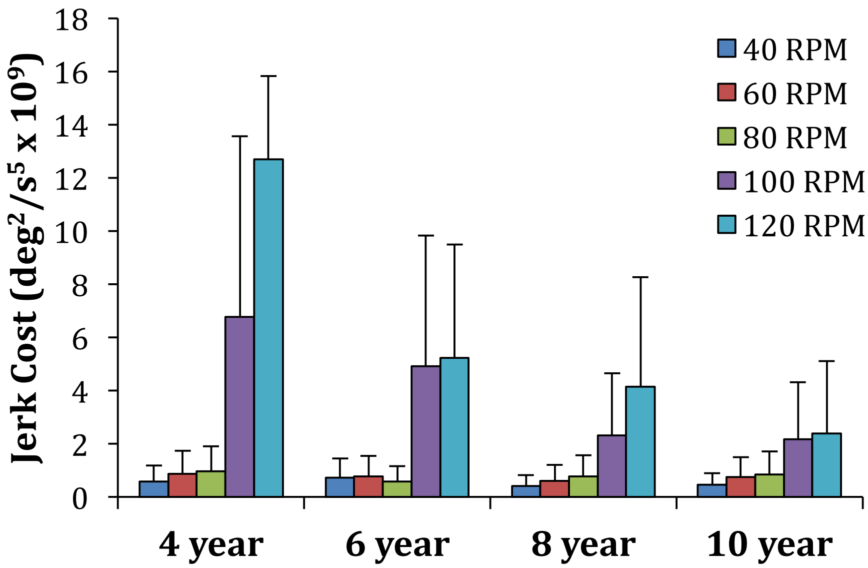


Figure 2.6: Average jerk cost for each group and each cadence.

DISCUSSION

In this study, we documented age-related changes in cycling on a stationary ergometer. Children pedal tricycles and bicycles, and it is a common activity of early childhood. Cycling is also becoming part of the therapeutic regimen. We undertook this study of the kinematics of the cycling task to contribute to a developmental timetable against which we can compare the cycling skills of children with disabilities.

Our work found that children as young as 4 years were able to cycle, but they were limited in their ability to maintain effectiveness across a broad range of task conditions (cadences). In comparison to their older peers, young children were less able to cycle at a predetermined steady state cadence as the target cadence increased. More trials were omitted in the youngest (4-year-olds) group than in the older groups (6-, 8- and 10-year-olds) at the higher cadences because the some of the 4-year-olds were unable to cycle at a steady-state at these cadences. Even though no statistical analysis was performed for drop out (there were too few participants for too many pairwise comparison tests), this result suggests that when task demand increases (by an increase in cadence), younger children are less able to adapt. This result is consistent with earlier findings in TD populations, where young children (5-7 years) deviated more from the set target cadence than older TD children (8-10 years) (Liu & Jensen, 2012).

We first were interested in the three-dimensional joint kinematics in TD children. Our results indicate that joint kinematics differ in 4-year-olds in comparison to the older children. We found that out-of-plane motion was affected, but no significant differences in the joint kinematics in the sagittal plane between the age groups were observed. The stability in the sagittal plane indicates that at a young age, coordination between the legs during cycling is already stable, as was found in other locomotors skills, for instance in walking and jumping (Jensen, Phillips, & Clark, 1994; Sutherland et al., 1980). However, this stability was not observed in the youngest age group concerning movement outside the plane of motion. In cycling, we found increased out-of-plane motion in 4-year-olds. This increased out-of-plane motion resembled the early onset of walking, where 1-year-old infants experience more external hip rotation and hip abduction than children of 2 or more years old (Ivanenko et al., 2004; Sutherland et al., 1980), and during running where young children initially run with a wide stride (Gallahue, Ozmun, & Goodway, 2011). Besides the increase in the base of support by widening the stride during gait and running, the increased hip rotation and abduction in locomotor skills can also indicate that the nervous system is exploring the potential motor solutions while learning a motor skill. Sutherland et al. (1980) found that 1-year-olds walked with slightly more external rotation of the foot than older children, while our cycling data showed more pronation in the youngest group. This difference is likely due to the restriction of a seated position with the feet attached to the pedal, forcing children to turn their feet inwards to compensate for hip abduction and rotation. The increased out-of-plane motion was not influenced by changes in speed, meaning that the 4-year-old children did not have increased out-of-plane motion in comparison to the older children as speed went up.

As children get older, they show more proficiency in complex motor skills (Hellebrandt, Rarick, Glassow, & Carns, 1961). Six-year-old children did not differ significantly from older children in cycling (8- and 10-year-olds), so it can be concluded that children are competent cyclists by the age of 6 years. In the frontal and transverse plane, children of 6-years and older cycled with their legs in a more neutral position than the 4-year-old children. They had less abduction in their hip and cycled with smaller pronation angles in the subtalar joint. These reduced angles indicate that the TD children "straighten" their legs, as they get older, which is similar to the development in motor control observed in waking (Sutherland et al., 1980).

Besides increased motion in the frontal and transverse plane, we were also interested in the development of movement fluency. The 4-year-old children who were able to cycle at high cadences, cycled less fluently than older children as cadence increased. All children exhibited an increase in jerk cost with an increase in cadence, however, the increase in jerk cost was significantly larger for the 4-year-olds than for the older children. This difference could be why some young children were unable to cycle at steady state at the higher cadences, as they reached their upper limits with regards to movement speed. The increased jerk cost and/or their lack of ability to cycle consistently at high cadences are both indications that their neuromuscular system lacked the capacity to adapt to higher task demands. In adult cycling, the activation of the muscles in the lower extremities occurs earlier in the revolution as speed increases (Neptune, Kautz, & Hull, 1997). Children younger than 7 years of age are unable to make proportional adjustments to their muscle activation pattern with an increase in cadence, even though children can produce high cadences up to 120 RPM (Chao et al., 2002). At higher movement speeds, children are unable to contract their muscles fast enough and this is likely the reason that the young children in the current study showed a reduction in cycling fluency at 100 and 120 RPM. This might be why not all of the youngest children were able to cycle at a steady state at the highest cadences.

Clinical relevance

Cycling is commonplace during childhood play, as a sporting activity (for example, junior triathlons), and as a therapeutic intervention. Our work suggests that cycling training in TD children younger than 6 years should be applied in moderation. The increase in jerk cost shows that 4-year-old children are not as adaptable to changes in task demands as are older children. Inefficient cycling performances can push children to their physiological limits because increased heart rate and oxygen levels are related to less fluent cycling motion (Fujii and Nagasaki, 1995). Furthermore, the altered kinematics in these young children can indicate an increased risk of long-term injuries due to repetitious strain in the joints and incorrect technique (DiFiori, 1999). A follow-up study is needed to identify the effects of such altered kinematics on joint torques in the frontal and transverse plane and to determine whether altered kinematics are a warning sign for high joint loadings that could lead to injuries.

We also found that inefficient cycling (less fluent cycling motion and increased out-of-plane joint motion) in young TD children is a normal feature of human motor development. In young atypically developing children inefficiency in cycling is often observed (Johnston et al., 2007; 2008b; Kaplan, 1995), and could actually be partly attributed to normal development. However, unlike TD children, atypically developing children fail to improve their cycling efficiency and do not move towards a sagittal planar motion as they age (Johnston, Barr, & Lee, 2008a). With increasing emphasis on early intervention, we have to take into consideration that performance variability is a consequence of early skill development. Clinical measures of progress, or the lack thereof, will need to be interpreted in the context of the developmental level of the client.

Summary

The results of this study suggest that 4-year-old TD children have altered neuromuscular control during cycling. They cycle with more out-of-plane motion and they perform cycling actions at high cadences with less efficiency than older children. By the age of 6 years, TD children straightened their movement patterns and followed a more fluent sagittal motion. These results show that even though young children can perform different cycling tasks, they use different biomechanical solutions to perform the tasks, especially as they approach their performance limits.

Chapter 3: Study 2

Kinematics and kinetics cycling: A comparison of young children with cerebral palsy and their typically developing peers

INTRODUCTION

Primary therapeutic goals frequently focus on mobility skills to help expand or maintain a patient's physical independence. Children with cerebral palsy (CP) are less physically active than their typically developing (TD) peers, as many have impaired locomotor skills due to spasticity and deformity in the legs (van den Berg-Emons et al., 1995; Wren, Rethlefsen, & Kay, 2005). Improving the level of physical activity of children with CP leads to an increase in muscle mass (Chen et al., 2012), bone density (Chen et al., 2013), aerobic capacity (Verschuren et al., 2007) and general health benefits such as a lower risk of coronary artery disease (Damiano, 2006). As well, cycling may be used as an alternative form of independent locomotion, as it is less dependent on balance compared to gait (Fowler, Knutson, et al., 2007a), and provides an exercise modality for healthy living (Mutton et al., 1997). For older children and adolescents with CP, cycling therapy has been shown to improve functional skills such as standing, walking and independent cycling (Williams & Pountney, 2007). As a therapeutic intervention, cycling is becoming more popular for children with CP, as there are many social, behavioral and health benefits associated with cycling. Few studies, however, have focused on the constraints induced by the task of cycling and the ability of children with CP to perform the task.

In research on children older than 10 years, those with spastic diplegic and quadriplegic forms of CP were shown to be capable of producing the cyclic movement, but they employed a different mechanical solution compared to TD peers (Johnston, Barr, & Lee, 2007; Johnston, Prosser, & Lee, 2008; Kaplan, 1995; Lauer, Johnston, Smith, & Lee, 2008; Rosecrance & Giuliani, 1991). Differences in muscle tone led to changes in movement trajectories and muscle involvement resulting in awkward cycling patterns compared to TD peers. Differences in kinematics and kinetics are a consequence of differential muscle activation because individuals with CP activate their muscles for a longer period in the crank cycle (Johnston et al., 2007; Kaplan, 1995) and have higher activation levels (Lauer et al., 2008). The difference in muscle activation increases disproportionally with an increase in movement speed, and there is significantly more co-contraction, thus joint stiffening (Johnston et al., 2007; Kaplan, 1995; Lauer et al., 2008). When propelling the crank, they spend a prolonged time pulling away from, rather than pushing on, the pedal (Johnston et al., 2008). Thus, while these older children and adolescents with CP are often capable of cycling a tricycle or ergometer, their movement solutions vary substantively from their TD peers.

As the emphasis on early intervention continues, the inclusion of a younger population is necessary to understand the early consequences of CP and the susceptibility of the child to benefits from intervention (Anderson, Spencer-Smith, & Wood, 2011; Landsman, 2006). From the literature on TD children, we know that there are significant developmental differences in the mechanics of cycling across the first decade of life (Jensen & Korff, 2004; Korff & Jensen, 2007) (Chapter 2). Children new to cycling (typically around 4 years of age) are more variable in the movement of their legs, they are less consistent in cycling cadence, and they show more out-of-plane motion and less cycling fluency than children aged 6-10 years (Chapter 2, (Jensen & Korff, 2004; Korff & Jensen, 2007). The cycling characteristics of children with CP are not unlike the behaviors of these young novice riders. While the young novice rider (e.g., 4-year-old) typically stabilizes their cycling movement quickly (by age 6), this is not the case for a child with CP. Of interest is whether an early, persistent intervention of cycling could be effective in improving and stabilizing the cycling movements of those with CP.

A persistent cycling regimen, however, must be designed for a specific goal, be that muscle strengthening (changes in resistance) or muscle endurance (duration and speed changes). In the therapeutic cycling research published to date, different regimens (e.g. increasing resistance or speed) have been employed (Fowler et al., 2010; Fowler, Knutson, et al., 2007a; Williams & Pountney, 2007). The weakness in this literature is that there is little evidence of standardized practices for cycling-based rehabilitation for CP. Although cycling training has resulted in positive effects, little is known about the cycling biomechanics of young children with CP and how these biomechanical features change in response to changes in the cycling regimen.

Also, because cycling protocols are designed around changes in resistance and changes in cadence, the workload for the child is altered during cycling training. Knowledge of the influences of these changes in workload on the cycling biomechanics are important, because children with CP have a greater risk of secondary injuries, such as pain, hip dislocation and subluxation, and patella stress fractures; especially if they are unable to walk independently (Lonstein & Beck, 1986; Murphy, 2009; Russo, Miller, Haan, Cameron, & Crotty, 2008; Samilson, Tsou, Aamoth, & Green, 1972). The risk of such injuries is increased by stress factors, such as repetition and high joint torques (Daffner, 1978; Piszczatowski, 2008). Importantly, the repetitive nature of a cycling task can lead to stress injuries if training intensities are not taken into consideration (Asplund & Pierre, 2004). Children with CP are, as well, less adaptable to increased task demands than TD peers (Fowler, Kolobe, et al., 2007b). Jerk cost as a measurement of motion fluency (Fujii & Nagasaki, 1995b; Schneider & Zernicke, 1989), can provide us with an indication of physiological demand (Fujii & Nagasaki, 1995a). It is a useful tool to distinguish between the abilities of different developmental groups (Chapter 2).

The understanding of the cycling biomechanics and the adaptability to changes in task demands on these cycling biomechanics can help design cycling therapies to optimize the benefits of treatment. Descriptive data regarding the cycling performances of children with different types of CP are needed to establish the range of performance diversity in this population before cycling training can be designed.

The goal of this study is to describe the cycling kinematics and kinetics of young children with CP. We are interested in how young children with CP cycle and how their performance changes with changes in task requirements. To create changes in the task requirements, we manipulated cadence, but not resistance. Young children with CP persistently demonstrate a low capacity for cycling against high resistance. Instead, we focus on cadence as a perturbation of child's ability to scale up the cyclic coordination of the lower extremities. We document the changes in kinematics and kinetics as cadence increases, and they approach their performance limits. This work will provide insights into the motor competencies of children with CP and add to the literature describing the capacity of children with CP to participate effectively in therapeutic cycling. Chronicling cadence dependent changes in the kinematics and kinetics will help us identify potential mechanical or physiological limitations in cycling regimens, with the goal of developing more effective therapeutic training in the future.

METHODS

Participants

Twenty-one children participated in this study. Fourteen children with different forms of spastic cerebral palsy were recruited. Their ages ranged between 5 and 7 years of age. Six of these children (GMFCS I-IV) were unable to cycle continuously on the upright ergometer without an ankle-foot orthosis or were unable to reach the pedals due to excessive hip adduction ("scissoring"), and one child could not sustain continuous cycling across multiple trials. These seven children were excluded from further studies. The remaining seven children (Table 3.1) were matched to 7 TD peers of the same age (within 2 months of age), gender, height, weight and cycling experience (Table 3.1). None of the participating children had received lower extremity surgery or suffered a traumatic fracture within the past 6 months. None of these participants had received botulinum toxin injections within the past 3 months, or undergone surgery for selective dorsal rhizotomy or placement of a Baclofen pump. The parent or guardian of each participant completed and signed an informed consent approved by The University of Texas Institutional Review Board and by the Institutional Review Board of Dell Children's Medical Center of Central Texas at Austin.

Participant preparation

Sixteen reflective markers (14 mm diameter) were affixed to the participant's body; at right and left head of second metatarsal, right and left calcaneus, right and left lateral malleolus, the upper third of the right tibia, the lower third of the left tibia, left and right lateral femoral condyle, the upper third of the right thigh, the lower third of the left thigh, right and left greater trochanter, right and left anterior superior iliac spine. Also, there were three markers aligned on each of the two pedals to track cycling movement.

The seat height and the crank length were adjusted to standardize the position of the participants on the ergometer. By adjusting the handlebars, the angle of the trunk with the horizontal plane was set to $60° \pm 3°$. The knee angle was set to $75° \pm 3°$ at top dead center (TDC) and $155° \pm 3°$ at bottom dead center (BDC) by adjusting the seat height and the crank length. The crank length was set to approximately 20% of the leg length. The participants were fixed to the bike with clipless pedals, and a backrest was attached to the ergometer, to give support to the torso to the children with CP (Figure 3.1). Children with CP did not wear any ankle-foot orthosis to enable the measurement of ankle motion during cycling. All participants were able to sit on the ergometer in this position before they started to cycle. The resistance was set for 5 W due to preliminary findings that young children with CP were not always able to perform at higher resistances than 5 W.

Figure 3.1: Ergometer with trunk support that was used during this study.

Protocol

Each parent or guardian filled out a questionnaire regarding the cycling experience of his or her child and to estimate the number of hours that they cycled in the past 5 years. The parents of the children with CP also filled out a questionnaire about the severity of their child's disorder and the motor skills they possessed. This information was used to determine the participant's position on the Gross Motor Function Classification Scale (GMFCS) (Alshryda & Wright, 2014).

Anthropometric measurements of each child were taken (height, weight, and leg length). After participant preparation, the participants were asked to cycle on a stationary cycle ergometer for 10-15 min to allow the participant to adjust to the test conditions and to warm up.

During data collection, the participants were asked to cycle at 30, 40, 50, 60, 70 and 80 revolutions per minute (RPM) at 5 W because children with CP were unable to cycle at higher cadences (Table 3.1). During preliminary data collection, children with CP had problems cycling at a resistance higher than 5 W; for this reason, we did not include higher resistances. For each trial, data were collected once the participant achieved steady state cycling. Steady state was defined as the moment the participant cycled at least 5 seconds around the desired target. The participants were required to stay within a range of plus and minus 4 RPM around the target cadence for the data to be included in the analysis. A 10-camera passive marker tracking system (Vicon Nexus 1.5.1) with residuals less than 0.3 mm was used to collect joint center positions during cycling at a sample rate of 120 Hz for 25 seconds. At the same time, force data were collected using custom-made pedals instrumented with two force transducers in each pedal (model 9251AQ01, Kistler, Winterthur, Switzerland). The reaction forces at the right and left pedals were collected at 1200 Hz. Feedback detailing the cadence was given on a display on the ergometer. Cycling conditions were performed in a random order, which was obtained in Matlab (7.0.1). In between each cycling trial the participant received, at least, 1-minute rest to avoid fatigue.

Data analysis

Data were processed using Matlab (7.0.1) and OpenSim (3.0). In Matlab, the position of the foot relative to the crank revolution was used to describe outcome parameters of the kinematic and kinetics. The kinetic data were low-pass filtered at a cutoff frequency of 20 Hz and the kinematic data at a cutoff frequency of 10 Hz. The kinetic data were down-sampled to 120 Hz to match the kinematic data.

The middle right pedal marker was used to determine the location of the crank angle, where 0° and 360° is TDC and 180° is BDC. This marker was used to calculate the speed of each revolution; a revolution was defined from TDC back again to TDC via a complete 360° rotation of the crank. Five consecutive revolutions within a range of +/- 4 RPM around the target cadence were used for analysis. If the participant did not remain cycling for five consecutive revolutions within the range, four or three revolutions were taken for further analysis (See Table 3.1 for successful cadences and the amount of revolutions taken per trial).

Crank angles were determined for each revolution using the middle wand marker on the right pedal. Jerk was calculated as the third derivative of the crank angle. Jerk cost for the extension phase (crank angle between 0° and 180°) and the flexion phase (crank angle between 180° and 360°) of the right leg were calculated using equation 3.1:

$$Jerk\ Cost = {}^{1}\!/_{2} \int_0^T J(t)^2 dt \qquad \text{(Eq. 3.1)}$$

where J(t) is the jerk and T is the motion time of the revolution.

Crank and pedal angles in the global reference frame were determined to transform the pedal forces from the reference frame to the global frame and the crank frame (into tangential and normal crank forces), using the middle and posterior wand marker of each pedal. Crank torque was determined by multiplying the tangential force with the length of the crank. The crank torque for each crank revolution was plotted, and the area underneath the curve was computed and was used as a measure of asymmetry. The amount of work delivered by the left leg was calculated in comparison to the right side by using equation 3.2:

$$\%\, Work\ Done = \left(\frac{Work\ Done_{left}}{(Work\ Done_{left} + Work\ Done_{right})}\right) \times 100\% \qquad \text{(Eq. 3.2)}$$

Joint powers were analyzed in the sagittal plane using the kinematics and the force pedal data. Starting at the ankle, from the net torque about the ankle joint, the torque due to the pedal forces was subtracted to determine the muscle torque about the ankle. This ankle muscle torque was multiplied by the ankle angular velocity to determine the ankle joint power. The ankle power was determined for 2 degrees of crank angle. A similar technique was used to determine the knee and hip power (Korff, Hunter, & Martin, 2009; Korff, Newstead, van Zandwijk, & Jensen, 2014). Extension and flexion phases were determined by the direction of the angular velocity in each particular joint. For each phase, the sum of power production was calculated.

Joint angles and torques were determined in the sagittal plane as well as in the frontal and transverse planes using OpenSim (Delp et al., 2007). An OpenSim musculoskeletal model was adjusted by van Zandwijk & Jensen (Chapter 2), to calculate joint motion in three dimensions in the left and right hip, knee and ankle. The scale tool in OpenSim was used, to scale body parts of the lower extremities using experimental marker data. Subsequently, inverse kinematics were performed in OpenSim, to estimate hip and knee flexion, adduction, and internal rotation, ankle flexion and subtalar angles.

Lastly, the inverse dynamics tool was used to find joint torques in the joints of the lower limbs during the cycling motion. For this tool, the pedal forces in the global reference frame were used. For each trial, the dependent measures from the revolutions were averaged. One of the assumptions of inverse dynamics in a cycling model is that the hips remain stationary (Fregly & Zajac, 1989). One child with CP (participant 4) was obviously rocking on the seat during cycling; his trials were taken out of the kinetic analysis. The 60-RPM trial of Participant 5 and the 30-RPM trial of Participant 6 were omitted for the same reason. The remaining children had small motions in the hip or had hip obliquity, which was assumed negligible for our inverse dynamics analysis.

The kinematic and kinetic parameters from the TD peers were averaged per cadence, and a 95 % confidence interval (CI) was calculated using equation 3.3 (Diekhoff, 1996):

$$95\%\ CI = \bar{X} \pm 1.96 \frac{\sigma}{\sqrt{N}} \qquad \text{(Eq. 3.3)}$$

$\bar{X}$ is the group mean, σ is the standard deviation and N is the number of participants. The children with CP were compared to the CI boundaries.

Table 3.1: Participants information.

Participant	Gender	Age	Type Spastic CP	GMFCS	Length (m)	Mass (kg)	Cycling Experience (hours)	RPM	Amount Revolutions Analyzed
1	F	5yr2m	Right Hemiplegic	I	1.09	17.7	34	30 40 50	3 5 4
		5yr1m			1.19	22.8	104		
2	F	5yr8m	Right Hemiplegic	I	1.13	15.9	40	60 70	5 5
		5yr7m			1.13	19.9	59		
3	M	7yr3m	Left Hemiplegic	II	1.27	23.6	15	30 40 50	3 5 5
		7yr3m			1.28	26.8	26		
4	M	7yr1m	Left Hemiplegic	II	1.21	34.8	10	40 50 60	4 5 5
		7yr2m			1.30	26.9	48		
5	F	5yr10m	Left Hemiplegic	I	1.06	14.3	10	40 60 70	4 5 5
		5yr11m			1.09	14.7	21		
6	M	6yr2m	Diplegic	III	1.13	19.3	50	30 50 60 80	5 5 5 5
		6yr0m			1.16	18.8	144		
7	M	7yr6m	Quadriplegic	IV	1.19	18.4	70	30 40 50 60 70 80	5 4 5 5 4 5
		7yr8m			1.26	23.4	176		

Note: Matched peers in the purple shaded column.

RESULTS

Fourteen children with spastic CP participated in this cycling study after parents indicated that their children were able to cycle. Seven children were excluded from the study because of an inability to perform the task. The remaining participants (GMFCS I-IV) were able to cycle at a steady state for several revolutions across multiple cadences (Table 3.1). No contractures were observed in any of the children with CP.

Clinical samples typically show high variability thus invalidating standard statistical analyses. The results presented here highlight two extreme cases (one child with hemiplegic CP and one child with diplegic CP). Select data from other participants will be presented when appropriate.. An overview of all participants and available data is shown in Table 3.2. Using the performance of the TD children, a 95% confidence interval was calculated for each variable. Table 3.2 notes those variables on which each CP participant exceeded the boundaries of the 95% CI.

Table 3.2: Biomechanical parameters for each participant.

Participant	Plantar flexion	Reduced plantar flexion motion	Hip or knee flexion	Joint angles frontal and transverse plane - hip	Jerk cost	Tangential force	Normal force	Shear force	% Work done	Joint torques sagital plane	Joint powers	Joint torques frontal and transverse plane
1	✓		✓	✓	✓	✓	✓	✓		✓	✓	✓
2	✓		✓	✓	✓	✓	✓	✓	✓		✓	✓
3		✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
4	✓		✓	✓		✓	✓	?	✓	?	?	?
5		✓	✓	✓	✓	✓	✓	?	✓		✓	?
6	✓		✓	✓	✓	✓	✓	✓		✓	✓	✓
7	✓		✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

Note: Tick marks indicate the parameters that exceeded the boundaries of the 95% CI. Question marks indicate the absence of data for the particular parameters.

How typically developing children cycle

How a child with cerebral palsy moves the crank through the cycle is best put in context with how a typical child moves the crank. Here we begin with the data on TD participants.

Joint angles TD peers

During the power stroke (the extension phase between 0º to 180º crank angle), TD children plantar flexed their ankle, and they dorsiflexed their ankle during the upstroke (Figure 3.2A). At the same time as the ankle plantar flexed, the knee and hip were extending (Figure 3.2B and 3.2C). The diagonal curve of the hip angle – ankle angle plot indicated that the hip and ankle coordinated in phase with each other, with reversals of motion at TDC and BDC. The crossing of the curve showed that the joint angles did not change at a constant ratio. The elliptical shape of the knee angle – ankle angle plot showed a smooth relationship between knee and ankle motion. The ankle started reversing the plantar flexion motion at BDC while the knee started to reverse the extension motion past BDC. Both joint reversed the flexion motion at TDC (Figure 3.3).

Furthermore, TD children cycled with hip abduction and internal rotation, and subtalar pronation (Table 3.3). In the knee, they cycled with knee internal and external rotation, as well as adduction and abduction. When the leg was flexing, the knee moved slightly lateral, and medial when the leg was extending.

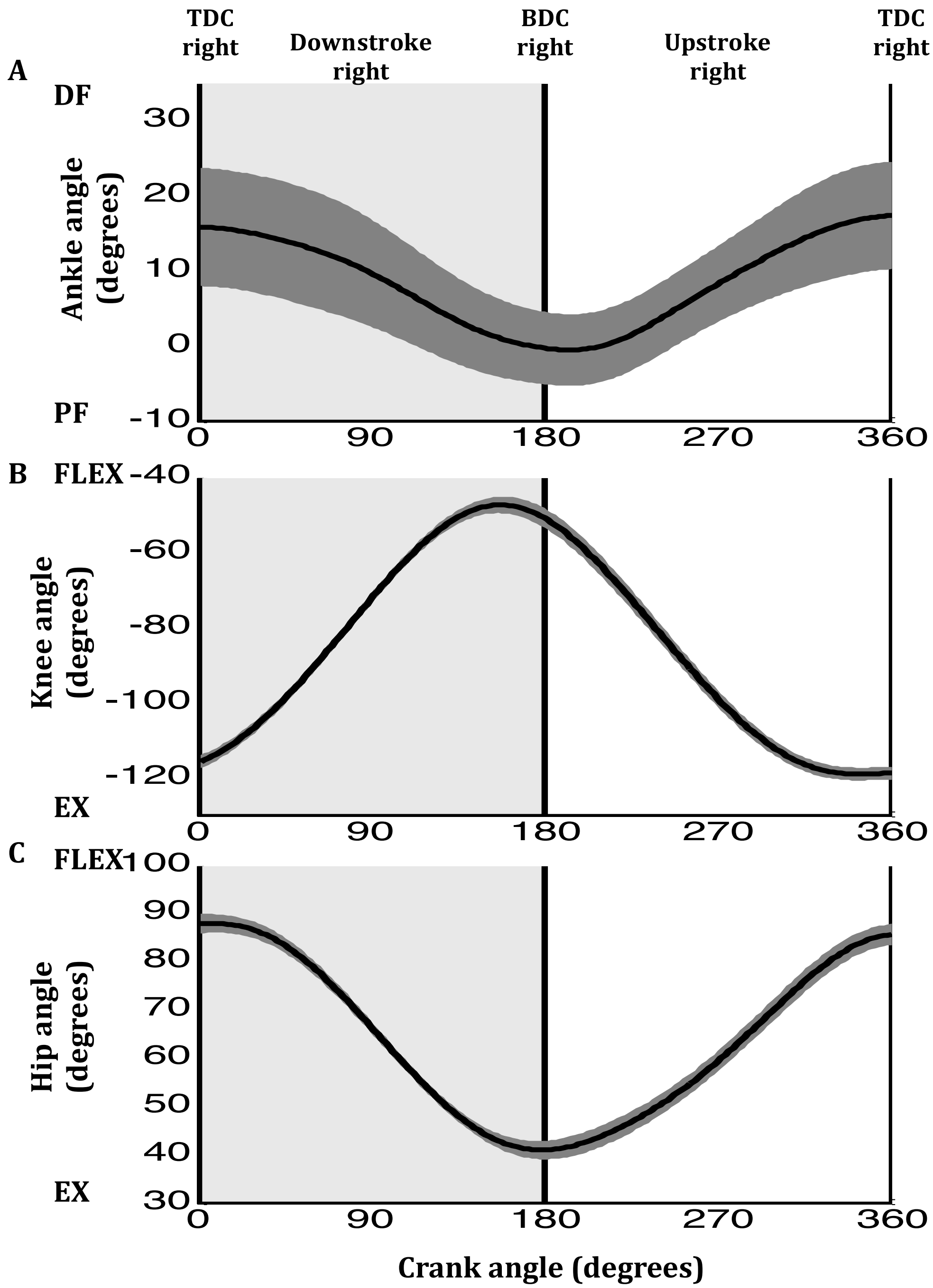


Figure 3.2 Joint angles in sagittal plane produced by TD peers. Right ankle (A), knee (B), and hip (C) kinematics during cycling at 60 RPM are displayed.

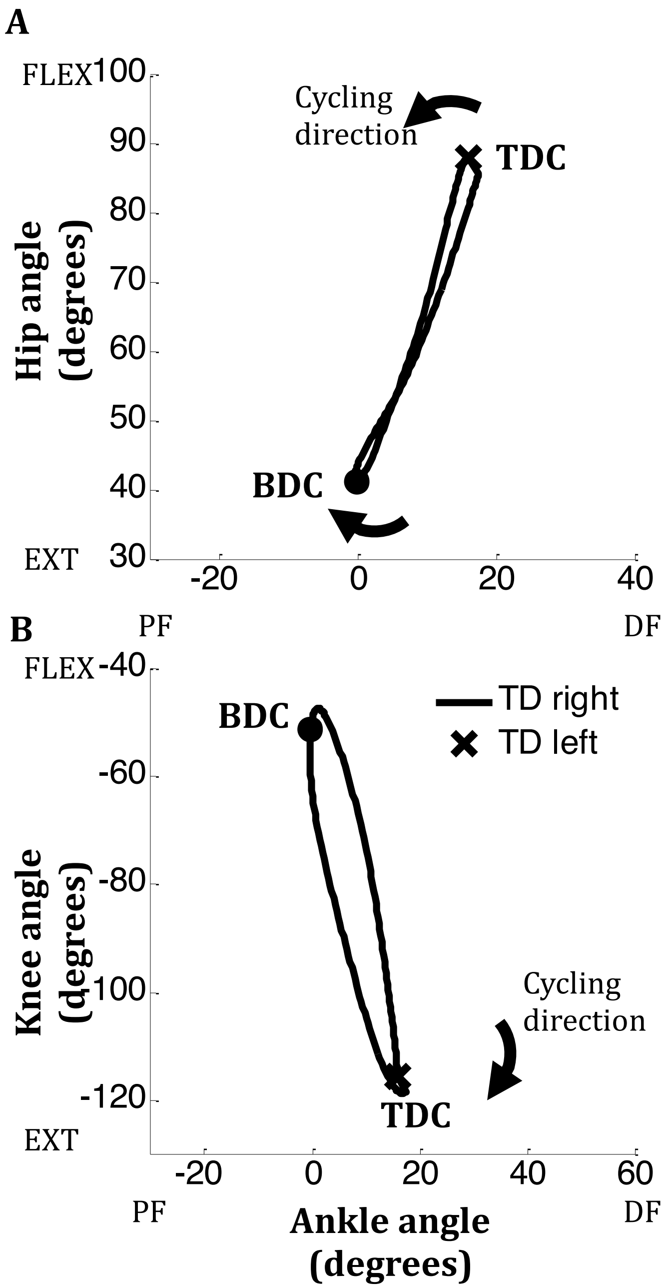


Figure 3.3: Angle-angle plots produced by TD peers. Hip-ankle angle (A) and knee-ankle angle (B) plots for the angles in the right leg of the TD peers during cycling at 60 RPM.

Table 3.3: Maximum and minimum joint angles and torques produced by TD peers for all joint motions.

Joint motion	**Minimum angle lower boundary 95% CI (°)**	**Maximum angle upper boundary 95% CI (°)**	**Minimum torque lower boundary 95% CI (Nm)**	**Maximum torque upper boundary 95% CI (Nm)**
Hip flexion/extension	43	88	-1	3
Hip ad/abduction	-12	-6	-1	0
Hip internal/external rotation	6	17	-1	3
Knee flexion/extension	-118	-47	-1	3
Knee ad/abduction	1	1	-1	1
Knee internal/external rotation	-4	7	0	-2
Ankle dorsi/plantar flexion	3	17	-2	2
Subtalar supination/supination	23	29	0	-1

Note: Positive angles indicate flexion, adduction, internal rotation, dorsiflexion, and pronation.

Jerk Cost TD peers

At the lowest four cadences, there was some variability in jerk cost between the cadences, but the jerk cost stayed below 1.0 deg^2/s^5 x 10^9 (below 1.2 deg^2/s^5 x 10^9 for the upper boundary of the 95% CI). Increase in jerk cost was observed when speed increased above 60 RPM (Figure 3.4).

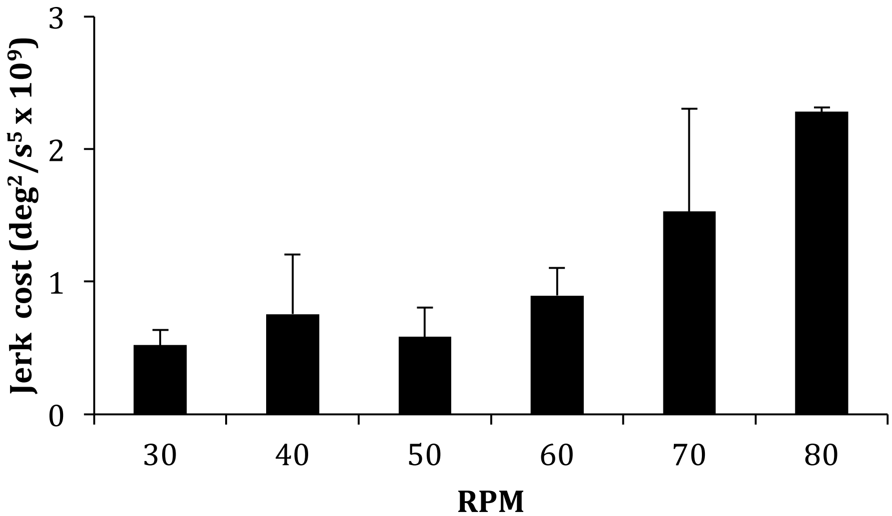


Figure 3.4: Jerk cost of TD peers at all cadences. The error bars represent the upper limit of the 95% CI.

Pedal forces TD peers

The TD children produced most of their tangential force during the downstroke, with the peak around 98º crank angle. The average peak tangential force over all six cadences ranged from 39 N to 50 N. The tangential force application was not symmetrical between both legs. The amount of work done by each leg was most asymmetrical at 30 RPM (ranging from 22% to 78 %) and least asymmetrical at 70 RPM (ranging from 41% to 59%, Table 3.4).

Around TDC, the TD children produced a downward force towards the crank center (normal force ranging between 24 N and 34 N). Around BDC, they produced a downward force that was away from the crank center (normal force ranging between -34 N and -48 N). Furthermore, these children produced during the downstroke an outward force by pushing sideways on the pedal away from the crank (ranging from 4 N to 8 N).

Table 3.4: Percentage work done.

Participant	**30 RPM** Percentage	**40 RPM** Percentage	**50 RPM** Percentage	**60 RPM** Percentage	**70 RPM** Percentage	**80 RPM** Percentage
TD	22-78	38-62	38-62	36-64	41-59	40-60
1	52	49	46	-	-	-
2	-	-	-	82	78	-
3	16	13	30	-	-	-
4	-	35	40	32	-	-
5	-	36	-	24	31	-
6	37	-	40	42	-	56
7	49	56	72	76	61	65

Note: Percentage work done by the left leg for each participant at different cadences (± 95% CI). The percentage work done by the TD peers was calculated as a deviation from 50% and then it was mirrored (i.e. flipped around the 50% mark) to find the lower and upper boundary of the 95% CI.

Joint kinetics TD peers

The highest joint torques produced by the TD peers were found in the sagittal plane (Table 3.3). The joint torques were not dependent on speed for the lowest five cadences but increased at 80 RPM. For example, between 30 and 70 RPM, the maximum hip extension joint torque was on average 6 N but increased to 11 N at 80 RPM. In the frontal and transverse plane, the TD children created small torques; abduction as well as adduction torque and internal as well as external torque.

Most of the power produced by the TD peers was produced during hip extension (Figure 3.5) while during hip flexion power was absorbed. Furthermore, power was generated during knee flexion and plantar flexion. At the lowest cadences, power was absorbed during knee extension. When the cycling speed increased, instead of absorbing power, the knee produced power during the extension phase. With an increase of power, less relative plantar flexion power was produced.

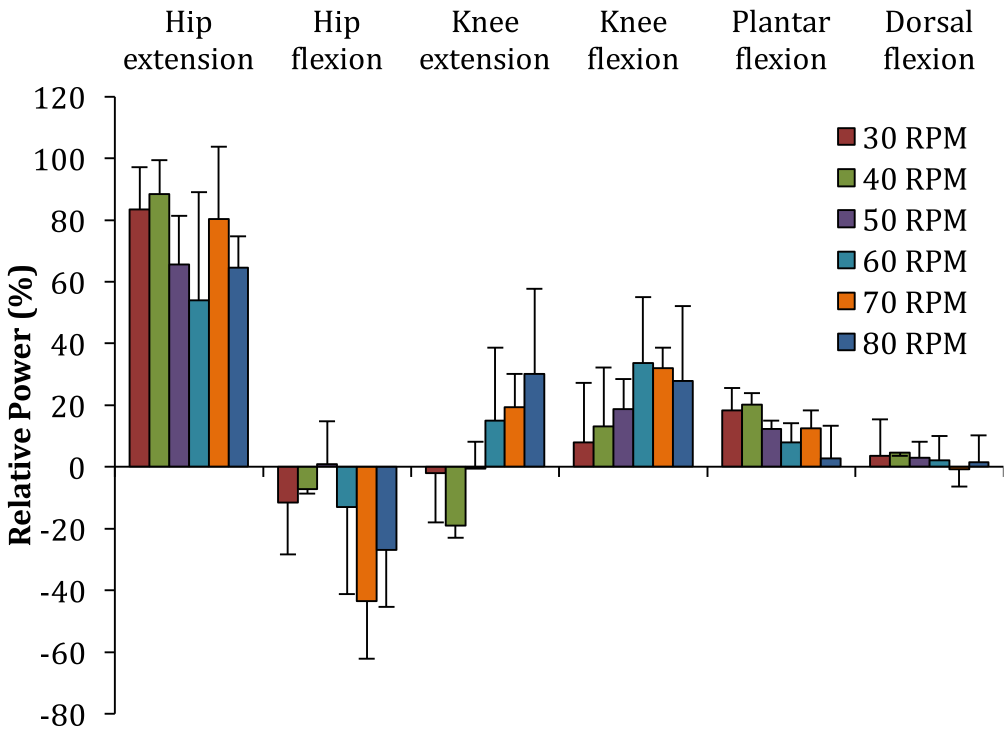


Figure 3.5: Relative joint power produced by TD peers for each cadence. Values from left and right sides were averaged.

Cycling as modified by the constraint of hemiplegic cerebral palsy

Hemiplegic CP

Five children with CP in this study suffered from spastic hemiplegic CP with low levels of involvement (GMFSC I-II) (Table 3.1). We were able to collect data detailing shear forces on the pedals for three of these children (Participants 1-3). The cycling performances of Participant 1 (CP 1) will be described in more detail to describe the overall differences with the TD peers. CP 1 was a female child with right hemiplegic CP and was able to walk in all indoor and outdoor settings. She had an upright bicycle at home but was not a regular cyclist. In the laboratory, she was able to cycle at three different cadences at steady state (30, 40 and 50 RPM).

Joint angles CP 1

Children with hemiplegic CP cycled with different joint angles in the sagittal plane, especially in the more affected leg. CP 1 cycled with her more affected ankle in a "toe-down" position. The plantar flexion angle fell outside the 95% CI of the TD peers. Due to this increased plantar flexion, a shift of the angle-angle trajectory of the right leg could be observed (Figure 3.6). In the less affected, left, leg, the trajectory of both the hip-ankle and knee-ankle angle plots were rounded in comparison to the elliptical shapes of the trajectories from the TD peers. This rounded shape was due to a phase offset (she plantar flexed and dorsi flexed her ankle, on average, 111^{o} crank angle later than her TD peers, which indicate decoupled coordination (Winstein & Garfinkel, 1989).

Besides different motions in the sagittal plane, CP 1 also had different joint angles in the frontal and transverse planes of both legs in comparison to the 95% CI of the TD peers (Figure 3.7). In the frontal plane, CP 1 had increased motion of the knee, in comparison to her TD peer (Figure 3.8). Especially her less affected knee, left, moved more medial-lateral than the TD peer. In her more affected leg, the path of the knee showed increased variability.

The differences in kinematics between CP 1 and the TD peers did not increase or decrease with increase in cadence.

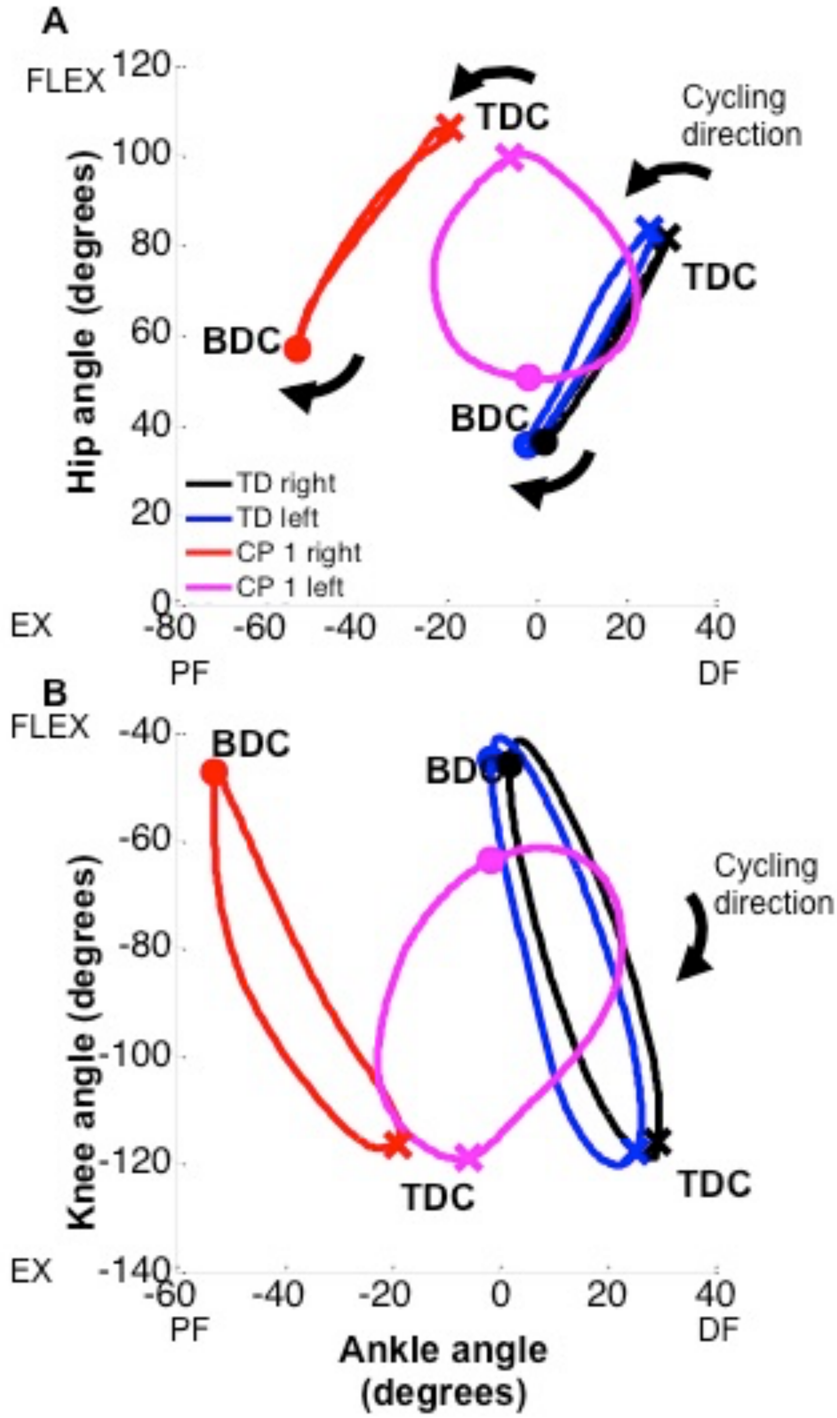


Figure 3.6: Angle-angle plots produced by CP 1. Hip-ankle angle (A) and knee-ankle angle (B) plots for CP 1 and TD peers during cycling at 40 RPM.

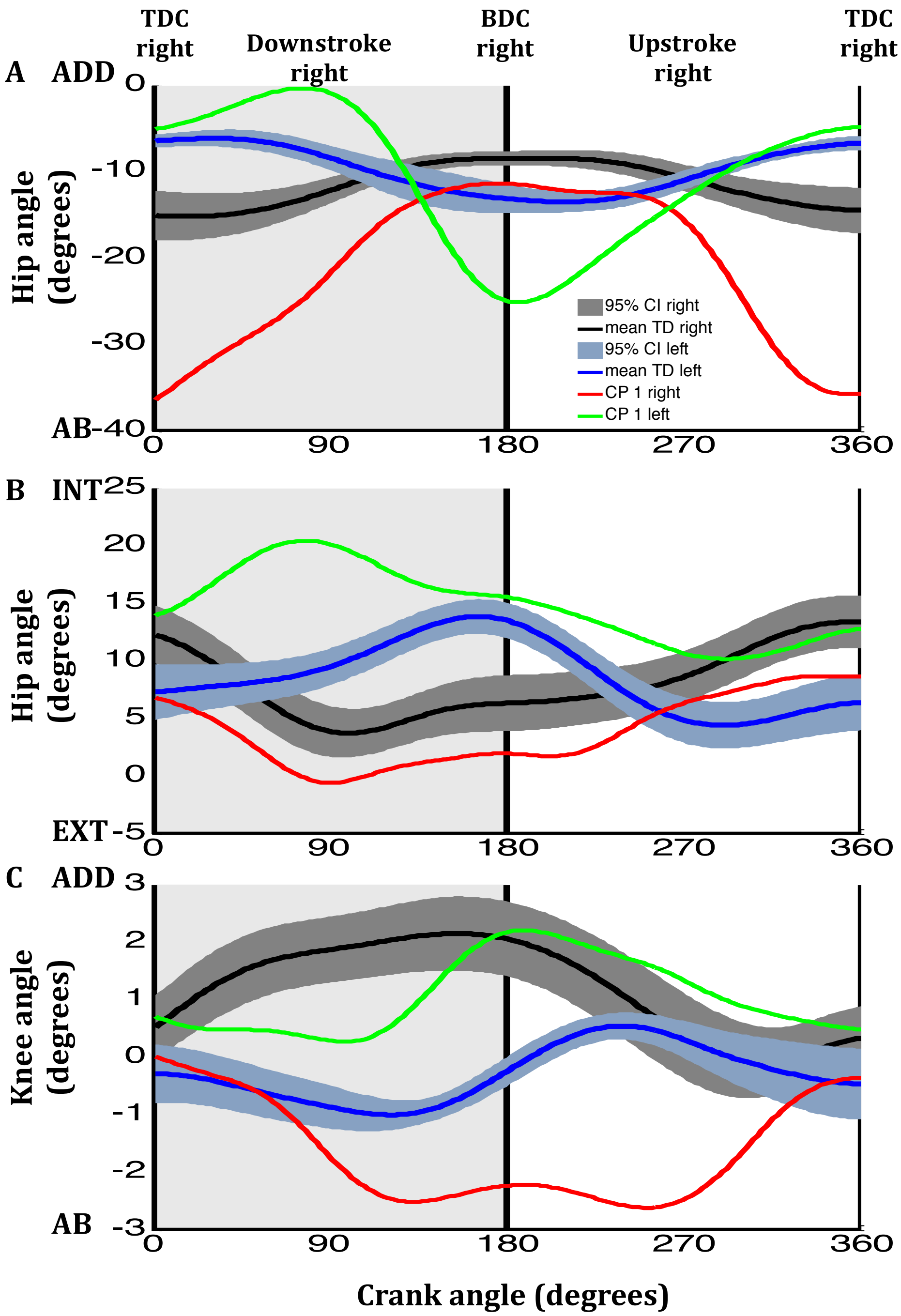


Figure 3.7: Joint angles in frontal and transverse planes for CP 1. Hip ab/adduction (A), hip rotation (B), knee ab/adduction (C) produced by CP 1 and the matched peers during cycling at 40 RPM.

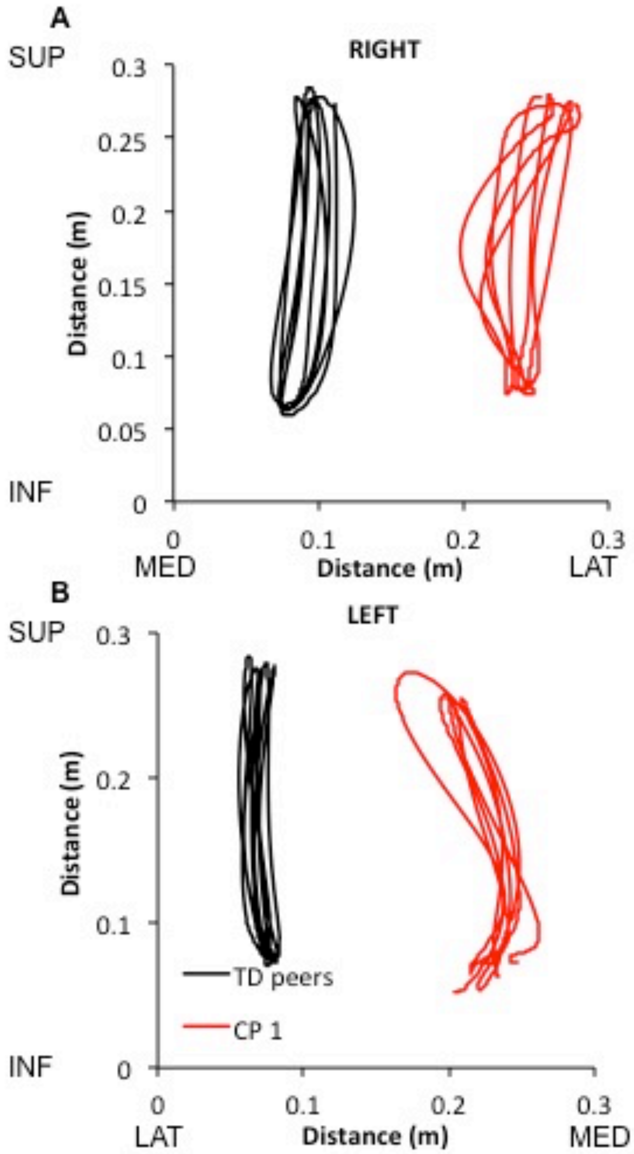


Figure 3.8: Knee motion in frontal plane for CP 1. Knee motion for CP 1 and TD peers during five consecutive revolutions in right (A) and left (B) leg observed in the frontal plane. Location of the knee is adjusted in the plot to put all for knee movements together. SUP = superior, INF = inferior, MED = medial, and LAT = lateral.

Jerk cost crank angle CP 1

Jerk cost showed that CP 1 cycled less fluently than the TD peers. The jerk cost increased exponentially with the increase in cadence in comparison to the TD peers (3.6 times larger than TD peers at 30 RPM and 10.0 times larger at 50 RPM) (Figure 3.9).

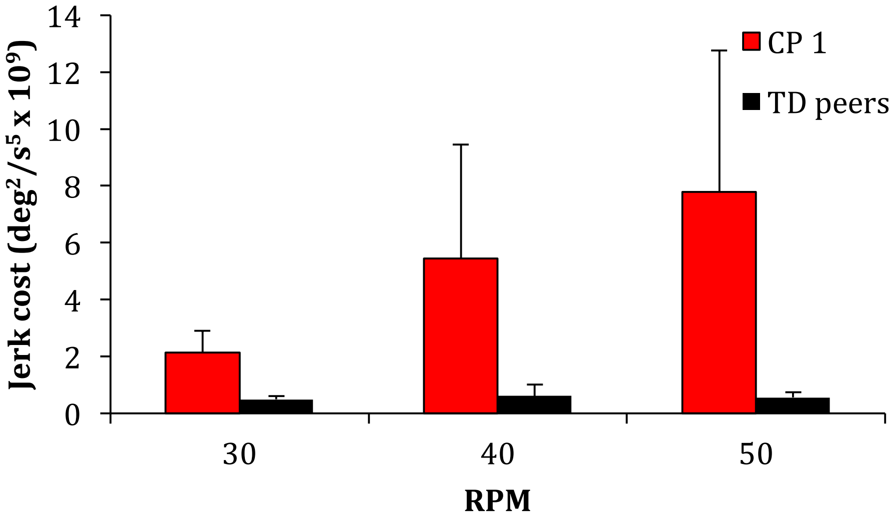


Figure 3.9: Jerk cost of CP 1 and the TD peers for three different cadences. The error bars for the CP 1 represent within-trial variability and the error bars for the TD peers represent the 95% confidence interval.

Pedal forces CP 1

Differences due to the changed kinematics in the sagittal plane could be found in the tangential (efficient) crank forces. With the right, more affected leg, CP 1 produced peak tangential force earlier (7º crank angle earlier) during the extension phase than the TD peers (Figure 3.10A). The less affected leg produced at all cadences an earlier drag on the pedal than her TD peers (average for all cadences 24º earlier) (Figure 3.10B). The early peak in drag (Figure 3.10A and 3.10B) coincides with the increased plantar flexion motion in the less affected ankle (Figure 3.6). CP 1 ceased dragging her feet during the upstroke, which helped the other leg by pulling the pedal up towards TDC. Around TDC of the more affected leg, CP 1 pushed downward with both legs. At this point, force was produced away from the crank center with the less affected foot at BDC and a force towards the crank with the more affected foot on TDC (Figure 3.10C). The normal force on the left less affected side was enlarged in comparison to the normal force production of the TD peers (on average 66 N enlarged). Even though the force application between both legs differed for CP 1, the amount of work done by each leg was similar to TD peers (within 95% CI) (Table 3.4). This amount of symmetry was not found in the other children with hemiplegia. The other four children produced more work with their less affected leg than with their more affected leg.

During the upstroke of CP 1's less affected leg, inwards forces were produced on the left pedal which were 6.6 N increased in comparison to the 95% CI interval of the TD peers (Figure 3.10D). Increased outwards forces during the downstroke of each leg were observed at both pedals. Outwards shear forces varied between cadences, but no linear relationship between changes in cadence and forces was observed.

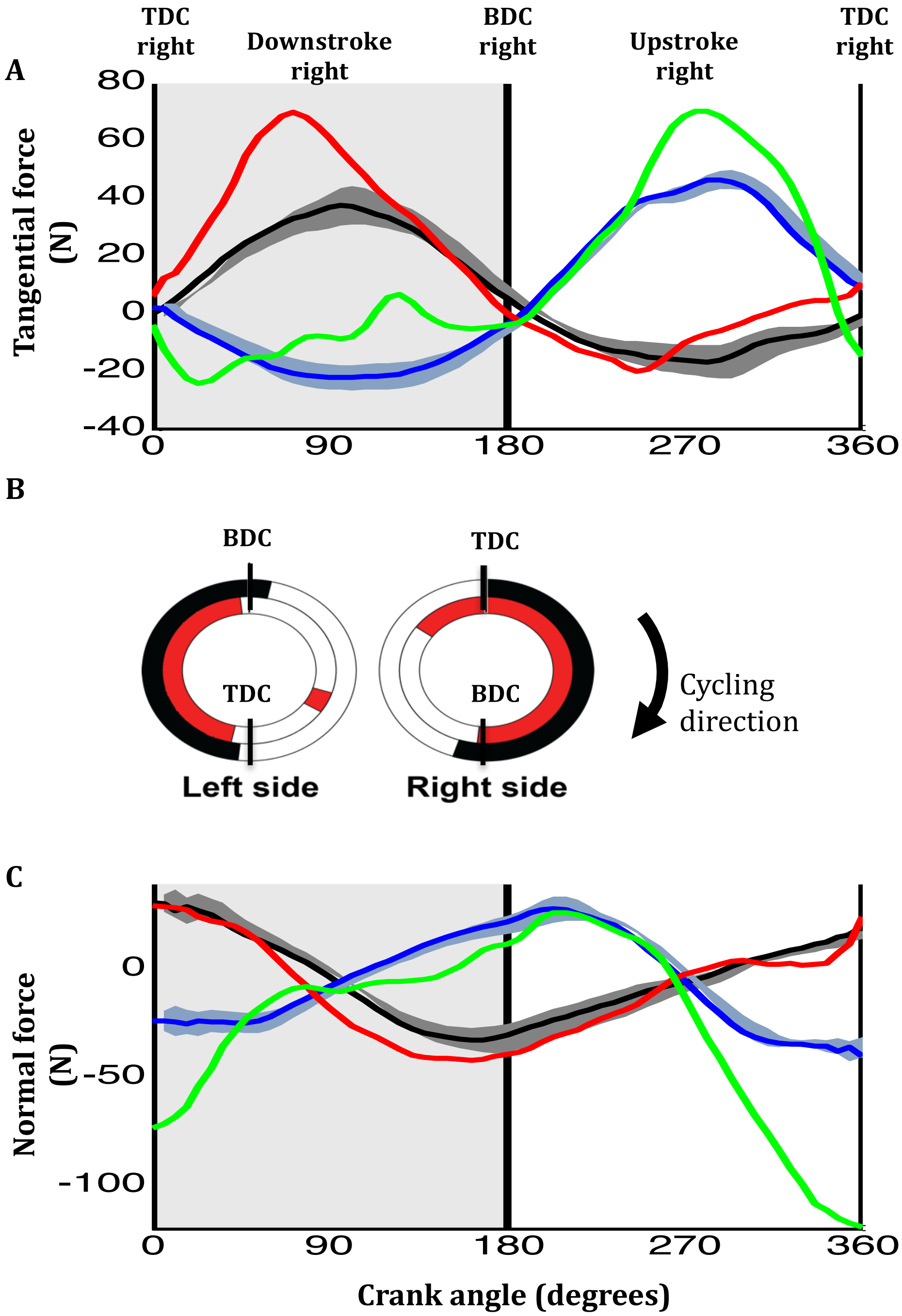

TDC
right
Downstroke
right
BDC
right
Upstroke
right
TDC
right
A
80
60
40
20
0
-20
-40
Tangential force
(N)
0
90
180
270
360
B
BDC
TDC
TDC
BDC
Left side
Right side
Cycling
direction
C
0
-50
-100
Normal force
(N)
0
90
180
270
360
Crank angle (degrees)

Figure 3.10

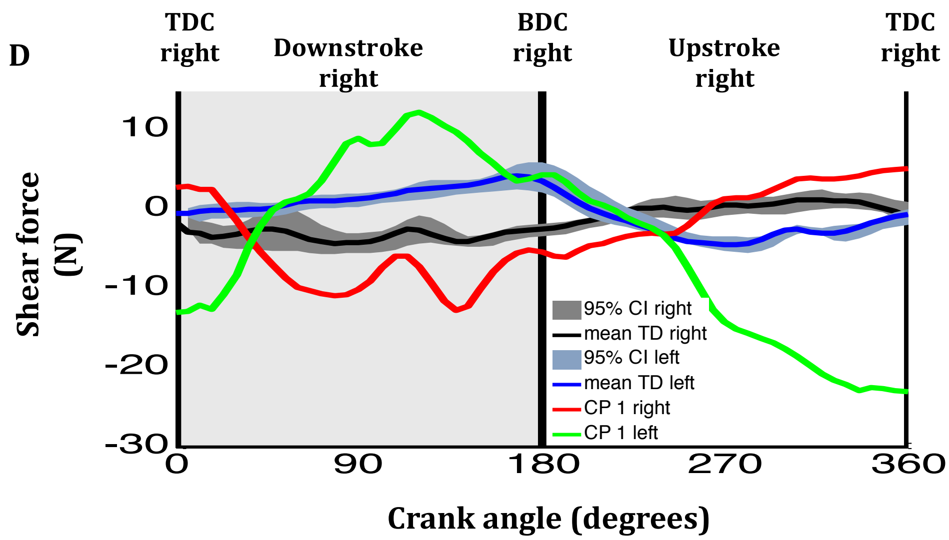


Figure 3.10: Crank forces from CP 1 and the TD peers at 40 RPM. Tangential forces (positive tangential forces accelerate the crank) (A), timing of application of positive tangential forces (propulsive force) on the pedal (B). The red circle is the positive force application by the participant 6 and the black circle the positive force application by TD. Normal forces (positive normal forces are forces towards the crank center) (C), and shear forces applied on the (D).

Joint kinetics CP 1

The kinematic and pedal force data related to differences in joint kinetics in the sagittal plane at both legs (Figure 3.11 and 3.12). Increased joint torques were observed in the sagittal plane of the more affected leg. At the hip of the more affected leg, the maximum flexion torque was on average increased by 1.6 times just before TDC. At the less affected hip, the maximum flexion torque was increased by 1.3 times at 30 RPM and the extension torque by 1.4 times at all cadences (Figure 3.11A). Increased knee extension torque was produced at all cadences during the extension phase of the more affected right leg (on average 1.4 times higher than TD peers) (Figure 3.11B). The less affected ankle also had increased plantar flexion torque around TDC (increased by 1.8 times the plantar flexion torque of the TD peers). All the joint torque in the frontal and transverse plane were larger than the 95 % CI, especially on the less affected side (Figure 3.12). The increased torque applications in the sagittal plane in combination with the angular velocity led to differences in the joint power productions (Figure 3.13). The TD peers produced most of the power for cycling in the hip; this happens during the extension phase. Children with CP, however, did not always produce most of their power during hip extension. CP 1 had an increased production of hip extension power in her more affected leg (on average 29% more power than TD peers) and produced more knee extension with her less affected leg (on average 321 N more than TD peers).

Although all children with hemiplegic CP had different torque and power productions (Table 3.2) an extreme power production was observed in the more affected leg of participant 3 (CP 3). With his more affected leg, CP 3 absorbed during the extension phase 393% from the total produced power at his knee. To compensate, he produced more than five times the pedal power by his hip. During the flexion phase the opposite occurred; the hip absorbed a large amount of power and the knee compensated for this absorption by producing power (Figure 3.14).

Similarities in kinematics and kinetics between all children with hemiplegic CP

The children with hemiplegic CP had increased ankle plantar flexion in their more affected leg, which led to alterations in hip angle – ankle angle and knee angle – ankle angle trajectories. The out-of-plane joint angles were affected on both the more and less affected sides. Pedal forces were affected in these children, as more forces were applied to the pedal with the less affected side in comparison to the more affected side and the TD peers. This asymmetry was also noticed in the joint torques in the frontal and transverse plane of these children. These participants also dragged their more affected leg earlier on the pedal. Four out of 5 participants produced less fluent cycling motion. When cadence went up, the fluency was more affected by spasticity.

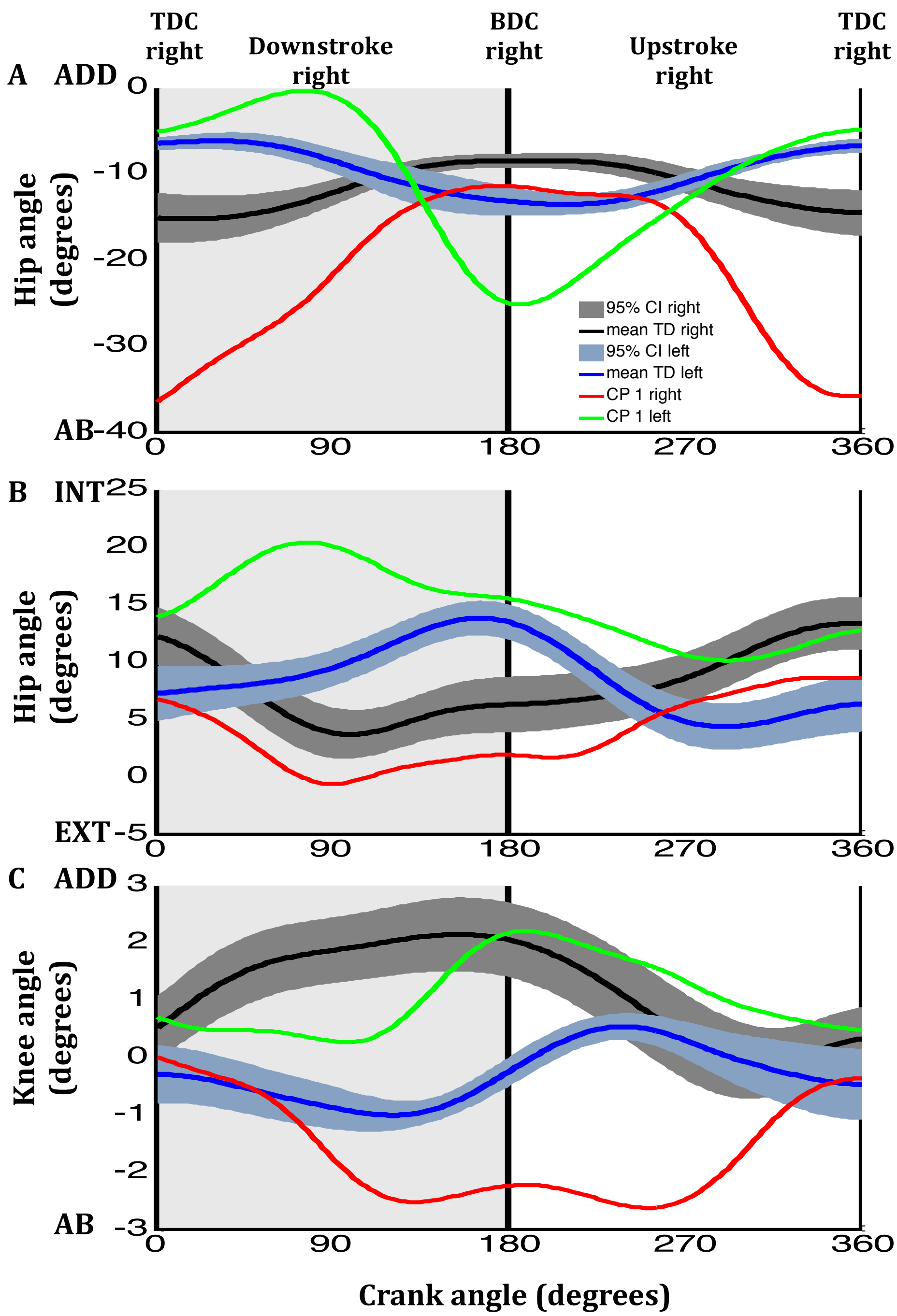
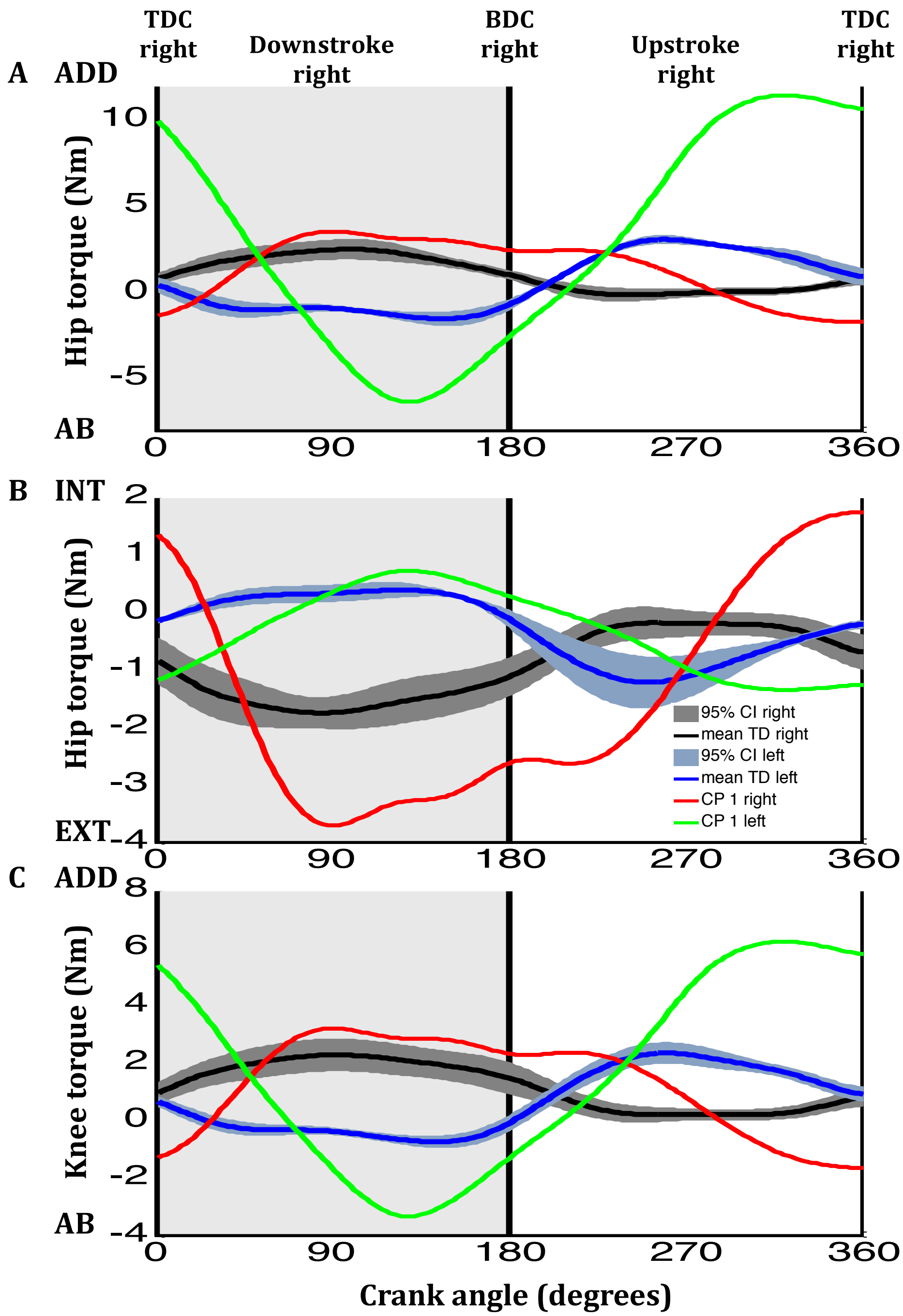


Figure 3.11: Joint torques in the sagittal plane for CP 1. Torques at left and right hip (A), knee (B) and ankle (C) for CP 1 and the TD peers during cycling at 40 RPM.



TDC
right
Downstroke
right
BDC
right
Upstroke
right
TDC
right
A ADD
10
5
0
-5
AB
Hip torque (Nm)
0
90
180
270
360
B INT
2
1
0
-1
-2
-3
EXT -4
Hip torque (Nm)
95% CI right
mean TD right
95% CI left
mean TD left
CP 1 right
CP 1 left
0
90
180
270
360
C ADD
8
6
4
2
0
-2
AB -4
Knee torque (Nm)
0
90
180
270
360
Crank angle (degrees)

Figure 3.12

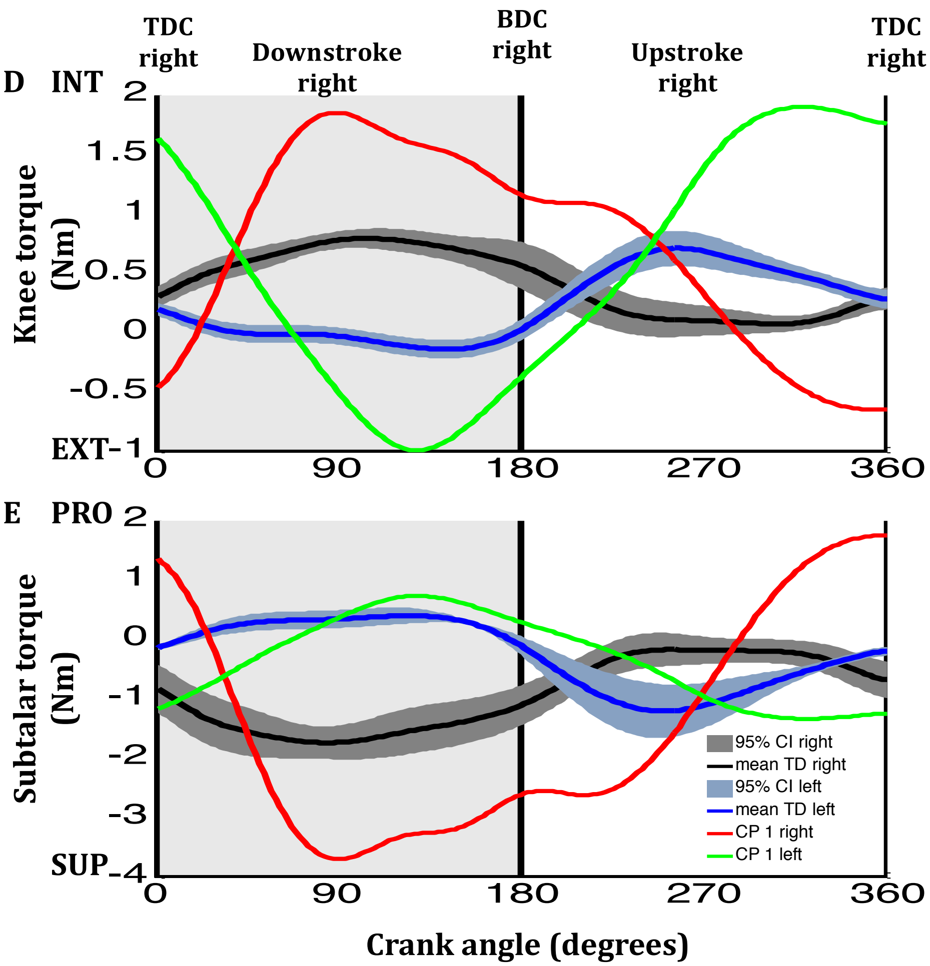


Figure 3.12: Joint torques in frontal and transverse planes for CP 1. Torques at left and right leg: hip ab/adduction (A), hip rotation (B), knee ab/adduction (C), knee rotation (D) and subtalar rotation (E) torques for CP 1 and the TD peers during cycling at 40 RPM.

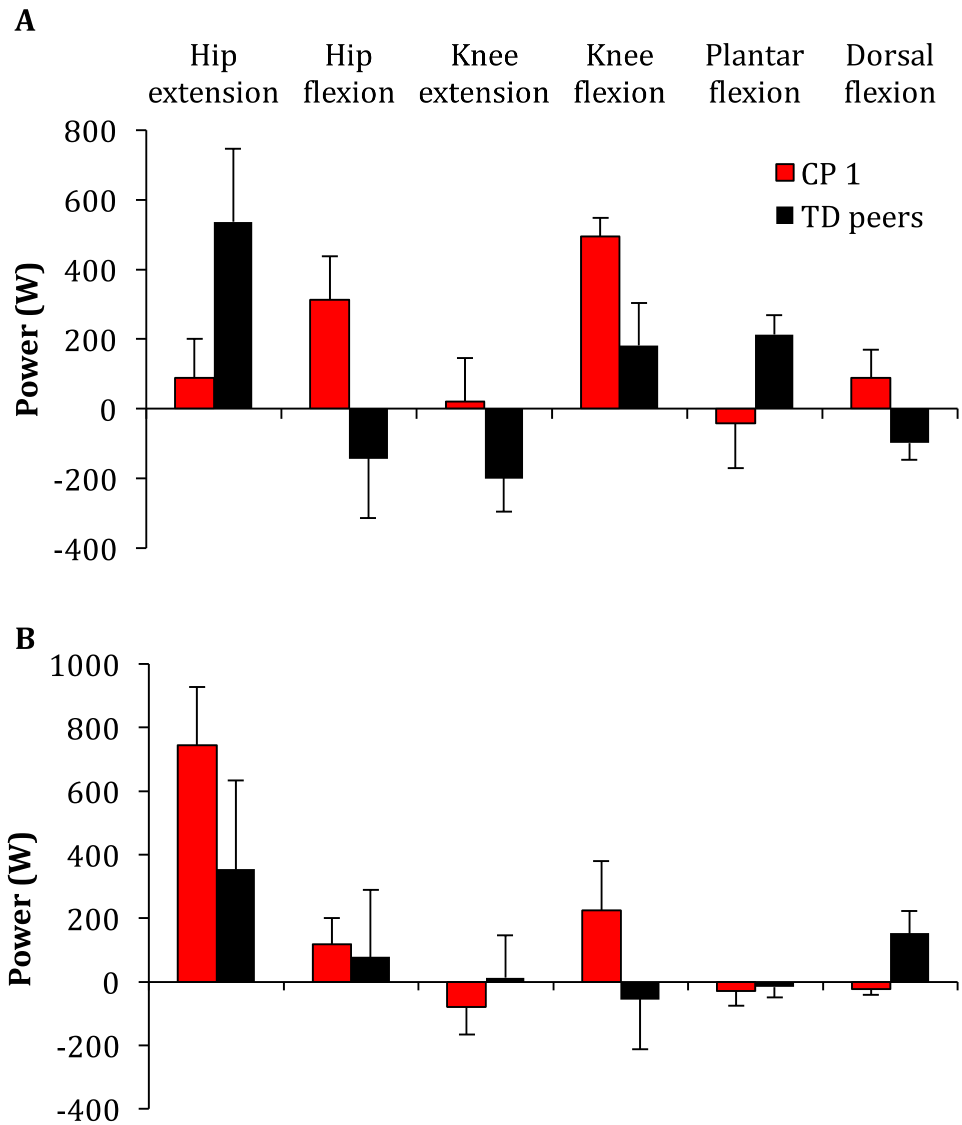


Figure 3.13: Joint powers in the sagittal plane produced by CP 1. Powers produced by CP 1 and the TD peers at 40 RPM in the left (A) and right (B) leg. The error bars for CP 1 represent within-trial variability and the error bars for the TD peers represent the 95% confidence interval.

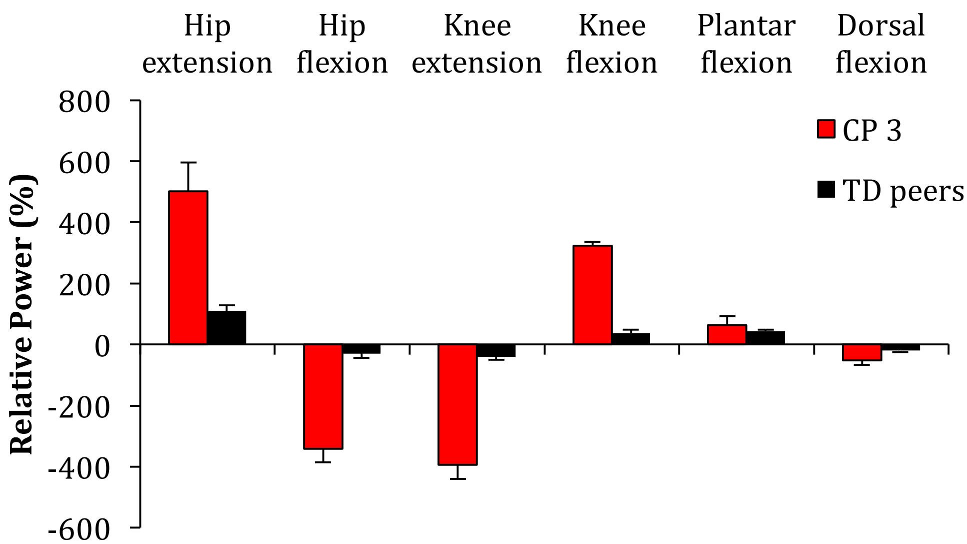


Figure 3.14: Relative joint power in the sagittal plane for CP 3. Powers produced in the more affected leg by CP 3 and the TD peers. The power is expressed as a percentage of the overall pedal power produced by the specific leg. The error bars for the CP 3 represent within trial variability and the error bars for the TD peers represent the 95% confidence interval

Cycling as modified by the constraint of diplegic and quadriplegic cerebral palsy

Two children that we studied suffered from spastic diplegic or quadriplegic CP, and had higher levels of involvement (GMFSC III-IV) than the hemiplegic CP children discussed above (Table 3.1). The performances of Participant 6 (CP 6) will be described extensively.

Participant 6

This participant, who had diplegic CP (Table 3.1), was able to walk with support, but was dependent on a wheelchair for mobility. The only cycling experience that he ever had was in a therapeutic setting.

Joint angles CP 6

CP 6 cycled with both feet in a "toe-down" position, hence a shift in angle-angle trajectories (Figure 3.15). Increase in cadence led to an increase in plantar flexion (on average between both ankles, there was an increase of 21º between 30 and 80 RPM) (Figure 3.16). Both the hip angle – ankle angle and the knee angle – ankle angle trajectories had a wide elliptical shape (Figure 3.15), which indicate that there was a decoupled coordination between the hip and ankle, as well as, a decoupled coordination between the knee and ankle.

CP 6 also had increased out-of-plane motion (Figure 3.17). Hip internal rotation and subtalar pronation in the right leg, and hip abduction and external rotation in the left leg were increased in comparison to the 95 % CI of the TD peers. The hip abduction and rotation increased with increase in cadence while the pronation was only increased at the lowest cadence. The differences in out-of-plane motion were best observed in tracking the knee motion in the frontal plane (Figure 3.19). CP 6 cycled with unpredictable knee motion (i.e., with high variability) while the knee of the TD peers followed a consistent path.

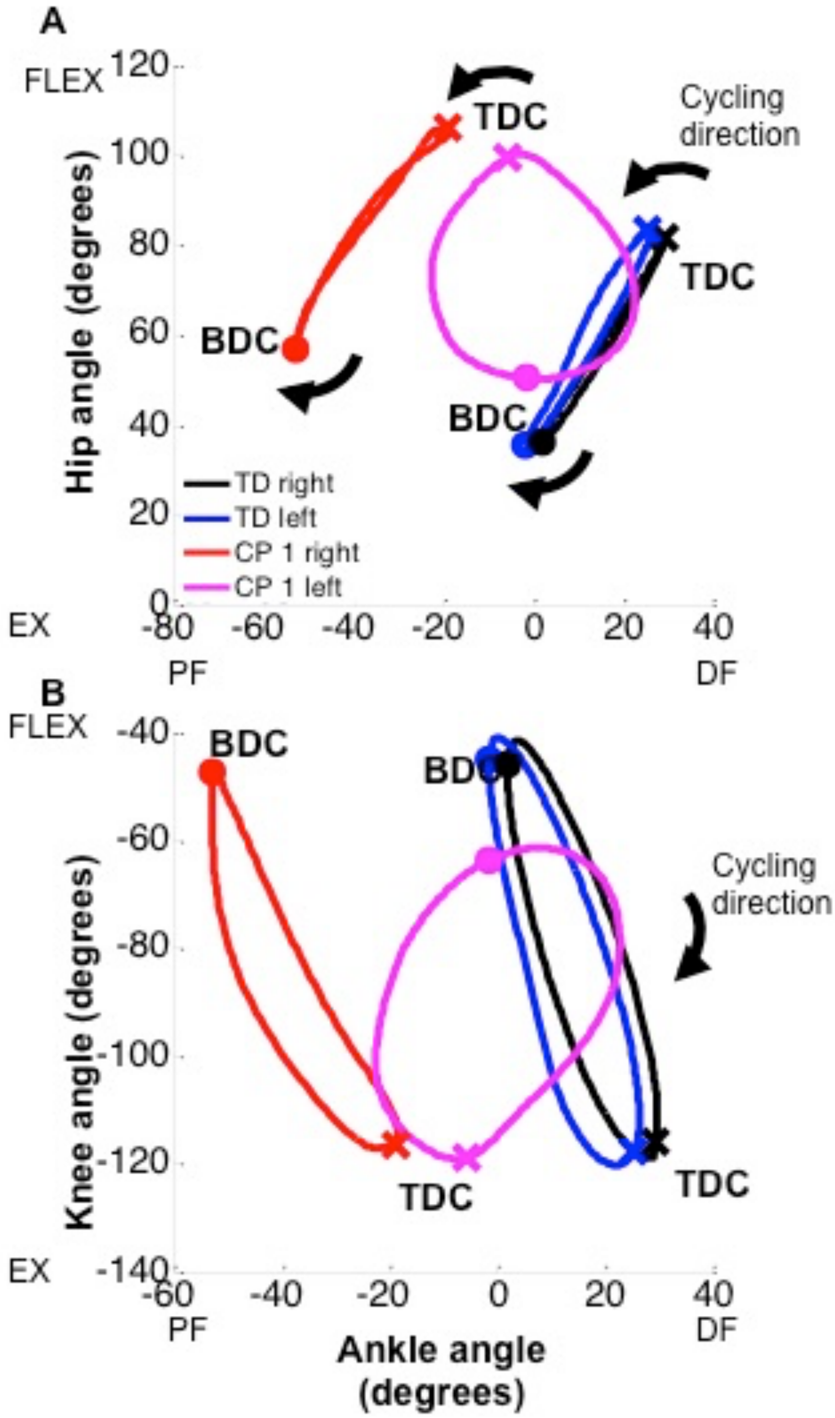


Figure 3.15: Angle-angle plots produced by CP 6. Hip-ankle angle (A) and knee-ankle angle (B) plots for CP 6 and the TD peers during cycling at 60 RPM.

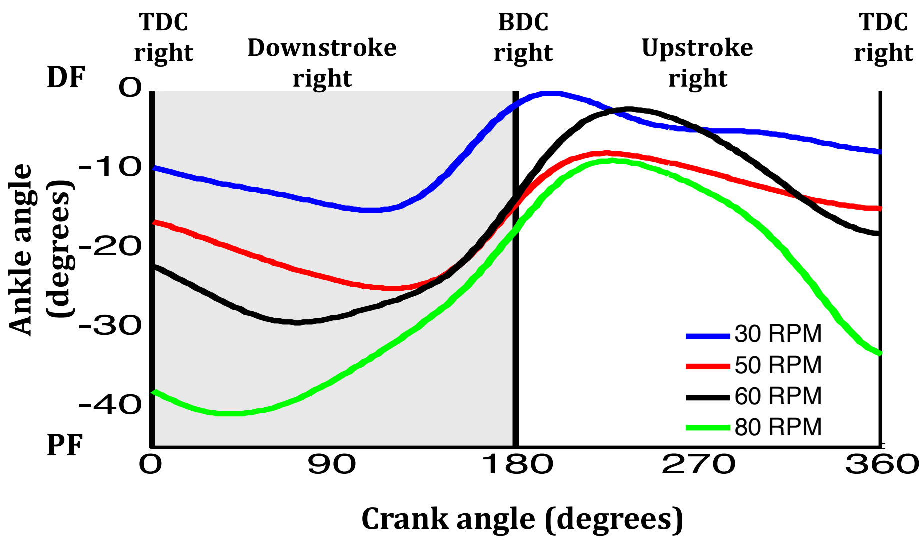


Figure 3.16: Ankle flexion at different cadences produced by CP 6.

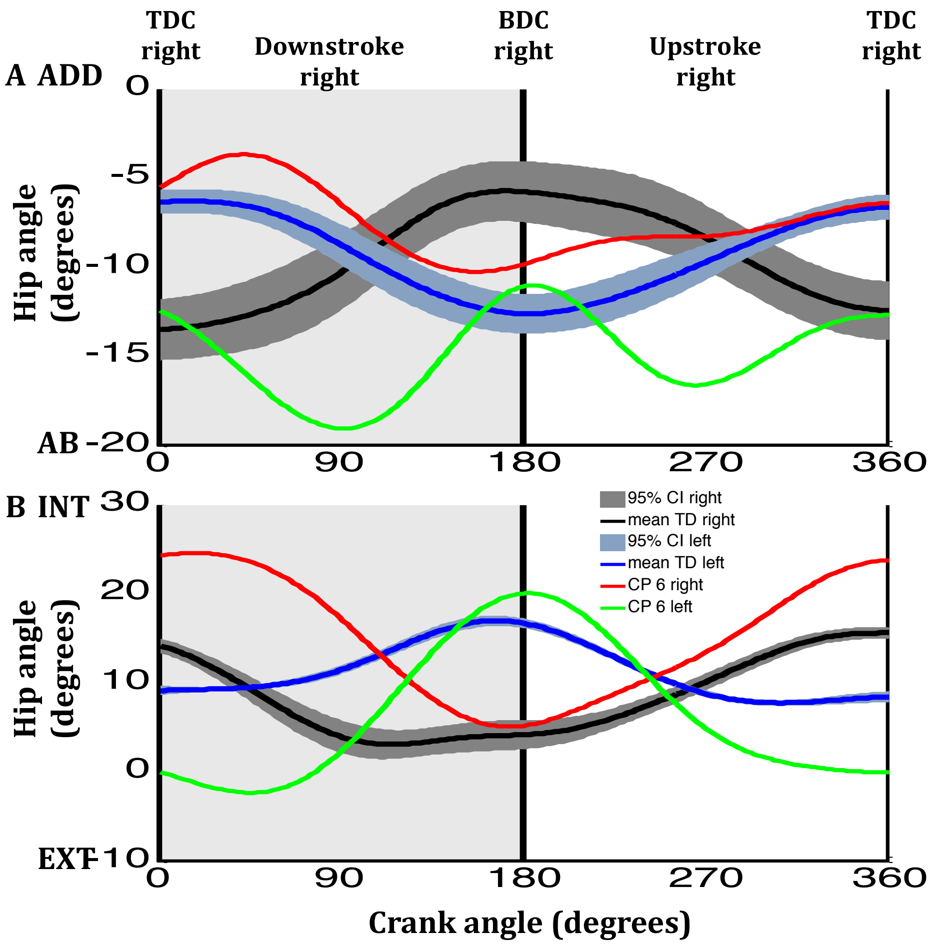


Figure 3.17: Joint angles in frontal and transverse planes for CP 6. Hip ab/adduction (A) and hip rotation (B) produced by CP 6 and the matched peers during cycling at 60 RPM.

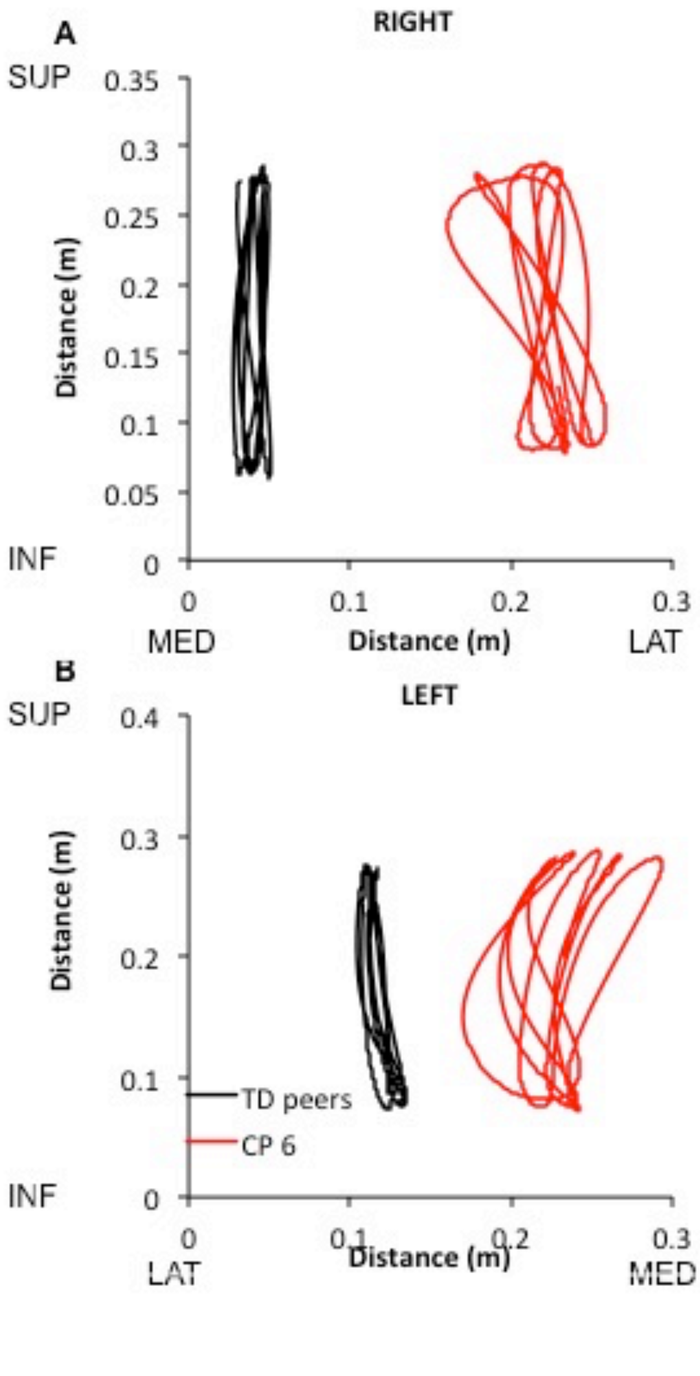


Figure 3.18: Knee motion in frontal plane for CP 6. Knee motion for CP 6 and TD peers during five consecutive revolutions in right (A) and left (B) leg observed in the frontal plane. Location of the knee is adjusted in the plot to put all for knee movements together. SUP = superior, INF = inferior, MED = medial, and LAT = lateral.

Jerk cost crank angle CP 6

CP 6 had increased jerk cost in comparison to the TD peers (on average 2.7 times higher). The jerk cost was influenced by cadence, implying that the jerk cost increased more rapidly with increase in cadence for a child with CP than for TD peers (2.5 $deg^2/s^5 \times 10^9$ more jerk cost than the TD peers at 30 RPM to 7.3 $deg^2/s^5 \times 10^9$ more jerk cost at 80 RPM, Figure 3.19).

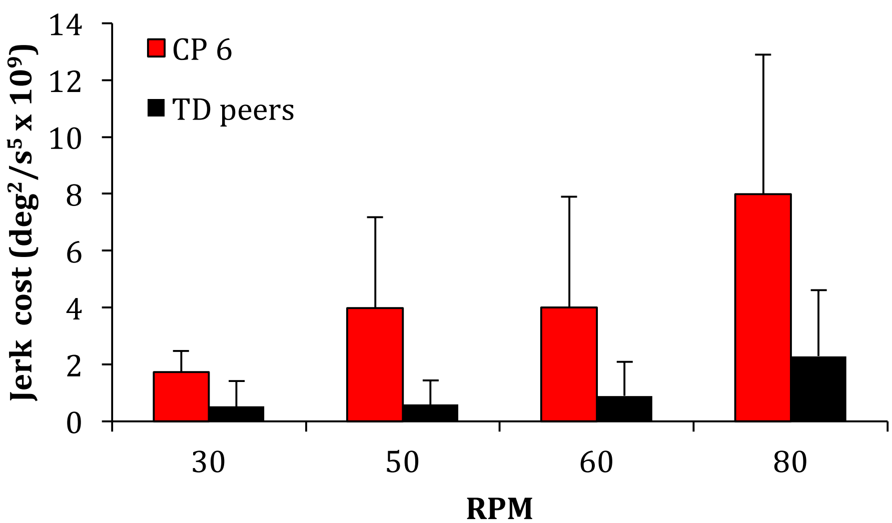


Figure 3.19: Jerk cost of CP 6 and the TD peers for four different cadences. The error bars for the CP 6 represent within-trial variability and the error bars for the TD peers represent the 95% confidence interval.

Pedal forces CP 6

CP 6 had earlier tangential peaks with his right leg compared to TD peers (average for all cadences 21º earlier) and started to drag both feet before BDC (Figure 3.20A) while the TD peers start dragging their feet after BDC (3.20B). The asymmetry that was observed in work done by each leg was similar to TD peers, considering the values fell within the 95% CI (Table 3.4). Looking at the normal forces, at 50 and 60 RPM he produced a downward force around BDC with his right leg, and at 60 RPM he produced a force away from the crank just before BDC of his left leg (Figure 3.20C).

At 60 RPM, CP 6 produced outward shear forces larger than the 95 % CI, at both pedals (Figure 3.20D). These forces were within the TD range at other cadences (Figure 3.20E). CP 6 produced inward forces on the pedal that fell outside the 95 % CI from the TD peers with his left leg, especially at 80 RPM.

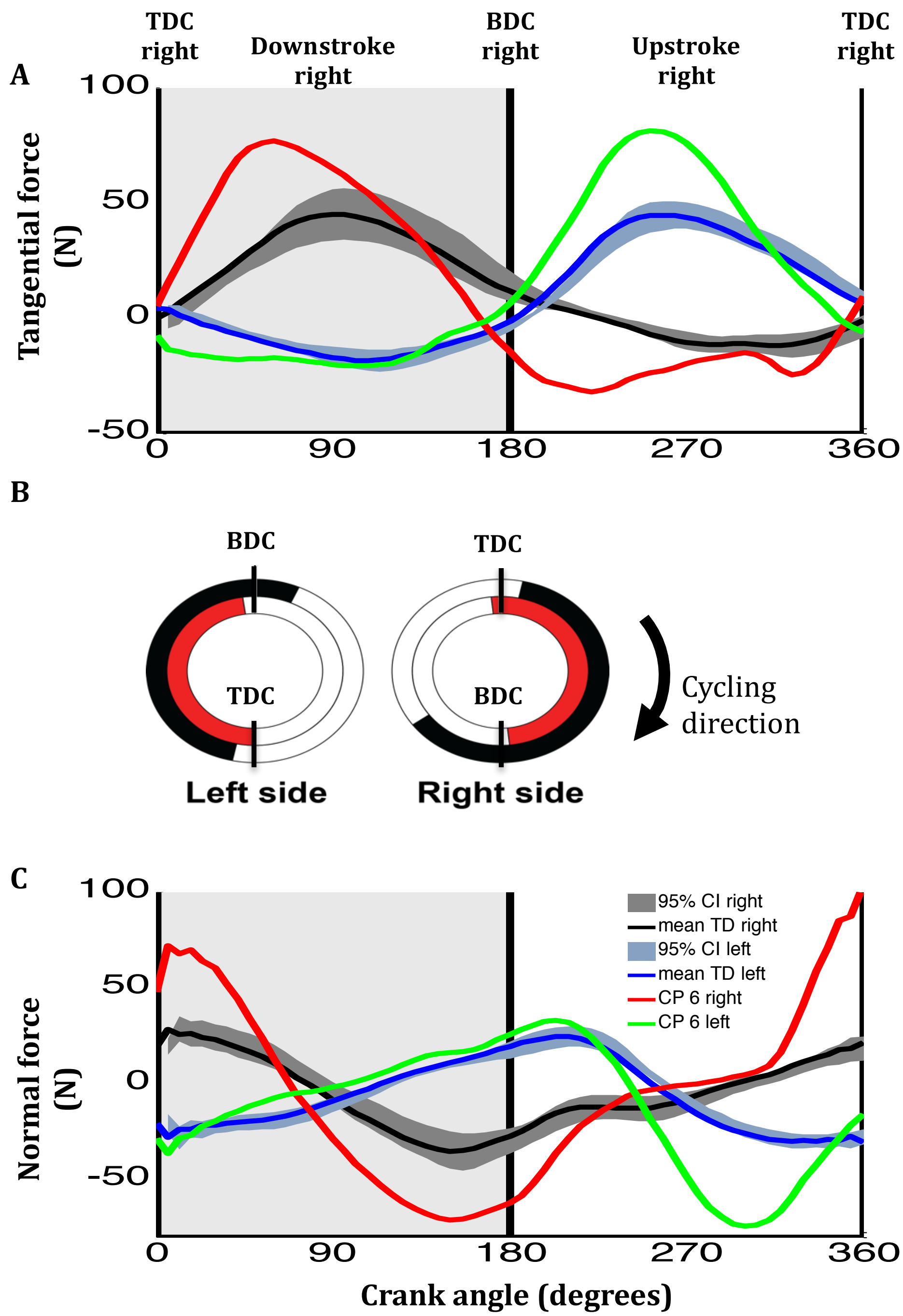

A
TDC right
Downstroke right
BDC right
Upstroke right
TDC right
Tangential force (N)
100
50
0
-50
0
90
180
270
360
B
BDC
TDC
TDC
BDC
Left side
Right side
Cycling direction
C
Normal force (N)
100
50
0
-50
0
90
180
270
360
95% CI right
mean TD right
95% CI left
mean TD left
CP 6 right
CP 6 left
Crank angle (degrees)

Figure 3.20

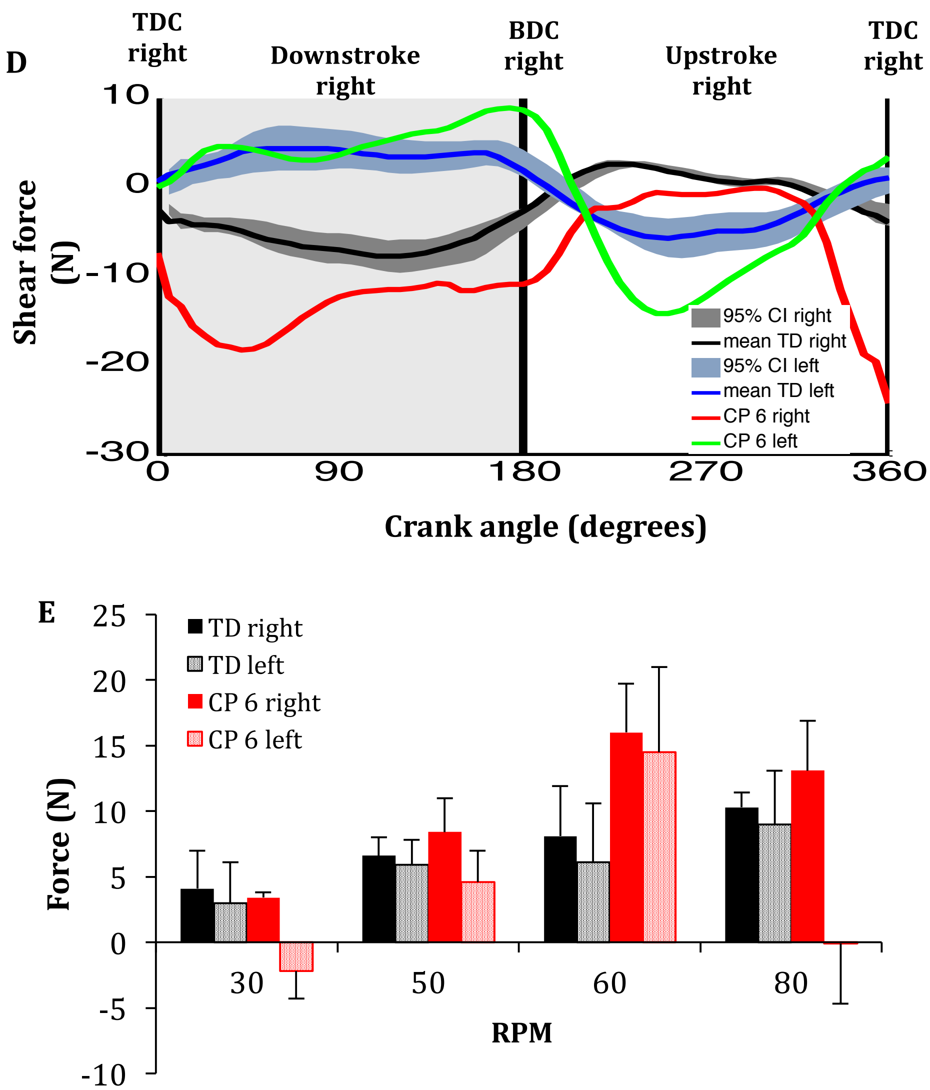


Figure 3.20: Crank forces from CP 6 and the TD peers at 60 RPM. Tangential forces (positive tangential forces accelerate the crank) (A), timing of application of positive tangential forces (propulsive force) on the pedal (B). The red circle is the positive force application by the participant 6 and the black circle the positive force application by TD. Normal forces (positive normal forces are forces towards the crank center) (C), shear forces applied on the (D), and maximal outward shear force at all cadences (B). The error bars for the CP 6 represent within-trial variability and the error bars for the TD peers represent the 95% confidence interval

Joint kinetics CP 6

At 50 and 60 RPM, CP 1 produced extension torque at the right hip around TDC, which contrasts TD peers that produce flexion torque around TDC (Figure 3.21A). Peak extension joint torques at both knees at 50, 60 and 80 RPM occured later for the child with CP in comparison to the TD peers (average for all cadences 13º crank angle later) (Figure 3.21B). At 60 and 80 RPM, at the left knee, these peaks for flexion were higher for the child with CP than his peers (2.2 times higher). In the frontal and transverse plane, adduction torque at the right hip, and hip rotation and knee adduction torque in each leg were larger than the 95 % CI of the TD peers (Figure 3.22).

The differences in joint torques were also observed in joint power. With the left leg, CP 6 produced less hip extension power (on average 23% less than the lower boundary of the 95% CI of the TD peers) and more knee extension power (on average 88.4% more than the upper boundary of the 95% CI of the TD peers) (Figure 3.23A). During the flexion phase, the muscles around the left knee absorbed power, while the TD peers produced knee flexion power. Even though he produced at 60 RPM more hip extension and knee flexion power with his right leg, on average between trials, his power production was within the 95% CI of the TD peers (Figure 3.23B). These differences in joint power were observed at all cadences.

Similarities in kinematics and kinetics between the children with diplegic and quadriplegic CP

Both children with di/quadriplegia had the joint angles, angle-angle trajectories, forces on the pedals and joint torques affected by spasticity in both lower extremities in all three planes of motion. Participants did not appear to favor either leg to apply force to the crank as was observed for the children with hemiplegic CP. All the biomechanical parameters were altered in both participants, and this worsened as cadence increased. Furthermore, more variability in biomechanics was observed between the trials in comparison to the children with hemiplegic CP and the TD peers.

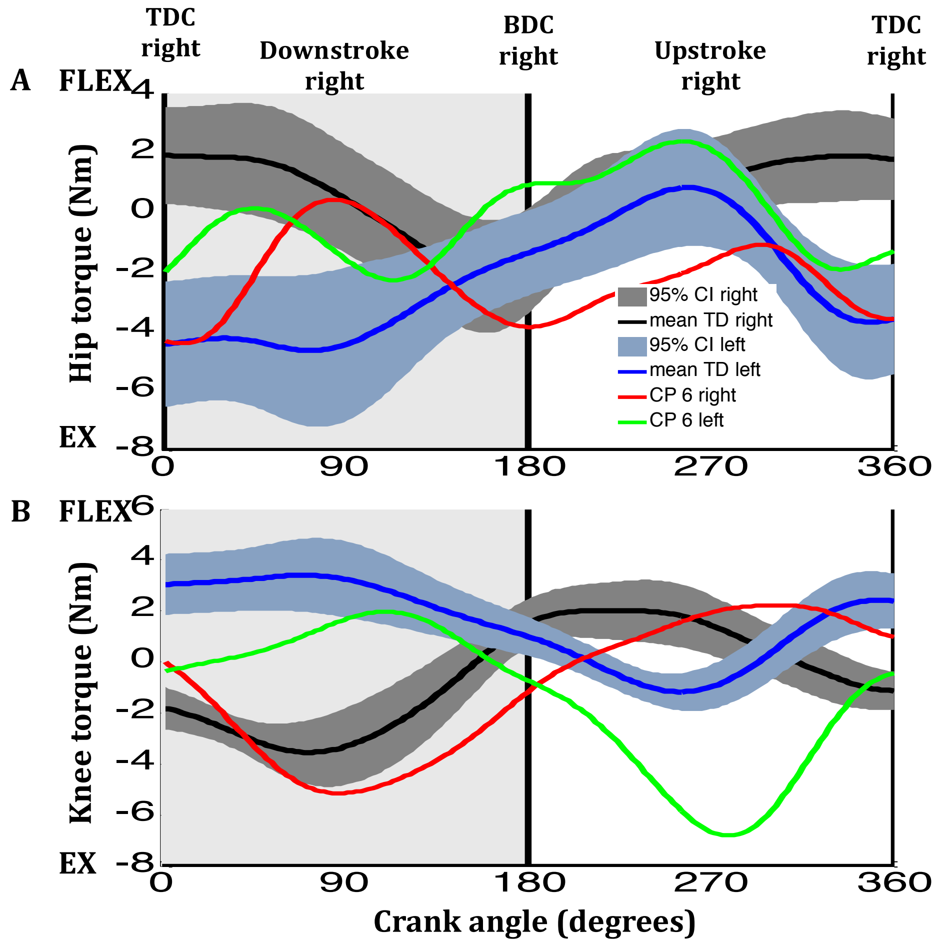


Figure 3.21: Joint torques in the sagittal plane for CP 6. Torques at left and right hip (A), and knee (B) for CP 6 and the TD peers during cycling at 60 RPM.

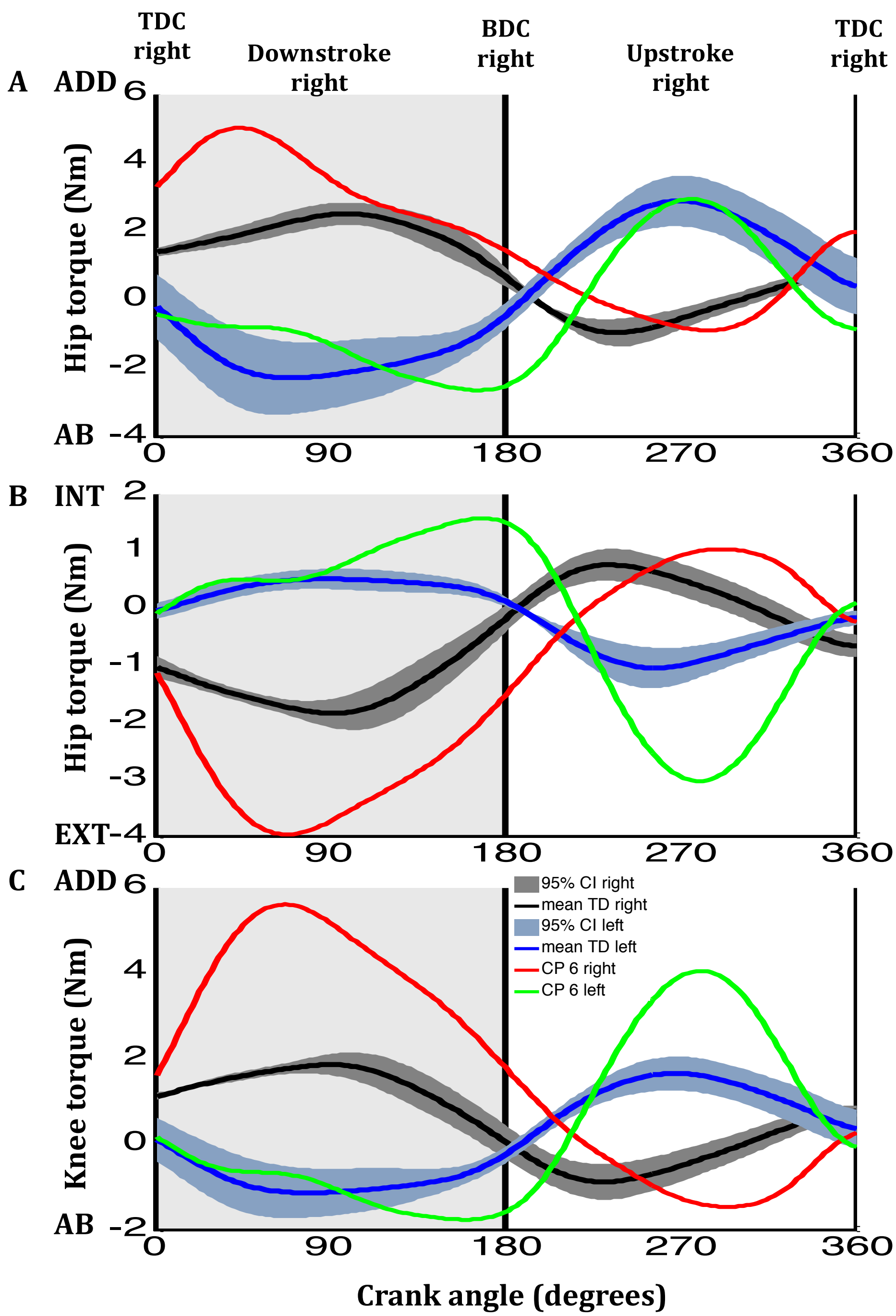


Figure 3.22: Joint torques in frontal and transverse planes for CP 6. Torques at left and right leg: hip ab/adduction (A), hip rotation (B), and knee ab/adduction (C) torques for CP 6 and the TD peers during cycling at 60 RPM.

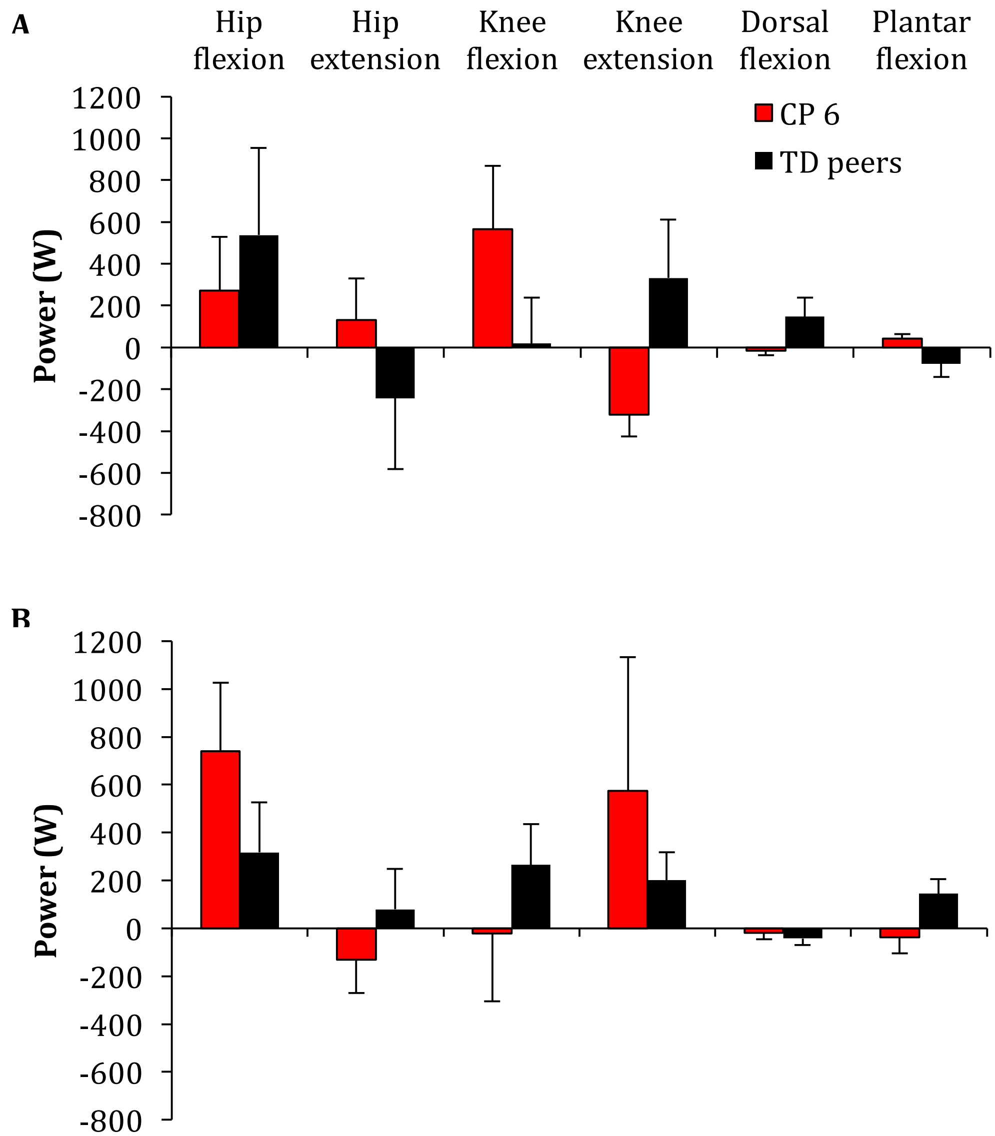


Figure 3.23: Joint powers in the sagittal plane produced by CP 6. Powers produced by CP 6 and the TD peers at 46 RPM in the left (A) and right (B) leg. The error bars for CP 1 represent within-trial variability and the error bars for the TD peers represent the 95% confidence interval.

DISCUSSION

The purpose of this study was to investigate the influence of spastic CP on the biomechanical cycling performances of young children and to provide recommendations for cycling therapy in children with CP. Children with CP performed similar cycling tasks as their TD peers but had different kinematic and kinetic solutions to these tasks. The children with CP were only able to cycle at a small range of cadences and cycled less fluently when task demand (i.e., cadence) increased.

Due to the standardized positioning of the ergometer, cycling tasks were similar for all the children that participated in this study. However, the performance of a cycling task can vary between individuals and within populations. For example, TD children cycle with increased joint motion (Chapter 2) and variability in angular velocity (Jensen & Korff, 2004; T. Liu & Jensen, 2012). In previous studies, it was established that TD children reduce their out-of-plane motion and primarily move in the sagittal plane as the children mature (Sutherland, Olshen, Cooper, & Woo, 1980) (Chapter 2). Young TD children (5-7 years old) cycled less consistently with the same crank angular velocity at a set cadence than older TD children (8-10 years old) and adults (T. Liu & Jensen, 2012).

When cycling speed was increased more differences between TD children and adults were observed. In TD children, it was observed that when cadences increased, the younger children were less able to perform a cycling task. In our previous study, we found that 4-year-old children struggled to cycle fluently at higher cadences, as they have a significantly larger increase in jerk cost between low cadences (40-80 RPM) and high cadences (100-120 RPM) than 6, 8 and 10-year-olds (Chapter 2).

Although young TD children cycle with a different technique and with more variability at cadences lower than 100 RPM compared to older TD children, they are still able to cycle. In our current study, we observed that young children with CP were able to cycle within a smaller cadence range and with a different technique than their TD peers. Even though the children with CP cycle differently, they still have the ability to cycle in recreational and therapeutic sessions, which increases the amount of repetition for this multisegmental motion. It is believed that performing these kinds of repetitive motions enhances motor learning because cycling is similar to gait a multi-segmental movement (Lauer et al., 2008). Since the children with CP cycle different than TD peers, cycling therapy can focus on changing the cycling motion to let the children perform a more "typical" locomotive skill, and train the major muscles that are responsible for "typical" motion. The current study highlights a few focus points for designing cycling therapy.

Plantar flexion leads to different pedal force application and joint kinetics in the sagittal plane

Firstly, differences in cycling performances were observed in the sagittal plane. The sagittal plane is the plane of motion of most locomotive skills. In this plane, the muscles accelerate the joint to produce forces and torques, resulting in the desired motion. In elite cycling, athletes use their uni-articular hip and knee muscles with the movement of the crank direction (e.g. the extensor muscles are activated during the extension phase) to generate power during the up- and downstroke. The bi-articular thigh muscles are used to switch from the downstroke and the upstroke and vice versa (Raasch

& Zajac, 1999). The power generated by the hip and knee muscles can contribute positively to crank acceleration by channeling power through the limb towards the ankle (Fregly & Zajac, 1989; Jansen, van Alfen, Geurts, & de Groot, 2013; Johnston et al., 2007; Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). The primary function of the ankle muscles are to position the foot appropriately to transfer the hip and knee power from the limb to the crank (Fowler, Knutson, et al., 2007a; Mutton et al., 1997; Neptune, Kautz, & Hull, 1997; Neptune, Kautz, & Zajac, 2000; Raasch & Zajac, 1999). In contrast to elite cyclists, young TD children are unable to use their muscles in an optimal manner to produce torques, which is thought to be due to a lack of capacity and less experience (Chao, Rabago, Korff, & Jensen, 2002; Korff & Jensen, 2007). When the muscles responsible for power generation during cycling are affected by spasticity, cycling performances in the sagittal plane was more affected in young children. Moreover, our data showed that young children with CP cycled with different foot angle, hip- and knee-ankle coordination, force application, and joint kinetics compared to young TD children.

Increased plantar flexion and/or reduced motion in the ankle angle were observed in the children with CP. With this increased plantar flexion, decoupled coordination between the ankle and the other joints in the lower extremities occurred. The increased joint kinematics with CP are likely the reason for changes in application of positive forces to the pedals. Plantar flexion motion during the downstroke is essential for creating tangential force because the soleus and gastrocnemius can transfer energy that is produced by the vastii and gluteus maximus to the crank by plantar flexing the foot (Zajac, 2002). If the ankle is locked in neutral position (zero degrees) a slight decrease in tangential peak, drag, and normal forces were found in TD adults (Pierson-Carey & Brown, 1997). When the ankle is in a plantar flexed position, less strength can be created by the plantar flexors, as they are shortened in this position (Zajac, 1989). The already weakened plantar flexors in individuals with CP leads to less force production by these muscles (Wiley & Damiano, 1998). Furthermore, children with CP drag their feet before the downstroke was completed, which causes a deceleration of the crank before BDC, making it harder to initiate the upstroke.

The joint kinetics that led to accelerations of the leg and crank were also different in children with CP. Increased peak joint torques and different joint powers were observed. Altered joint kinetics indicate that children with CP have a different strategy for developing the power to push the crank around. Changes in power production to meet the force requirements of cycling are not exclusively a feature of atypical development. Indeed, a reduction in hip power production was previously observed in children younger than 7 years of age (Korff & Jensen, 2007). This indicates that joint torque production is influenced by age and disability-related factors, but that the latter can lead to more changes than the former. The support of a dynamic ankle foot orthosis could be applied to improve the kinematics in the sagittal plane. This orthosis gives the child with CP the freedom to move the ankle enough for typical cycling motion, but it will reduce the ability to cycle in a plantar flexion position. It is expected that the reduced plantar flexion motion will lead to more typical cycling kinetics. However, further research is needed to investigate if the orthosis indeed would result in pedal forces and joint kinetics similar to the kinetics produced by TD peers.

Increased kinematics and kinetics in the frontal and transverse plane

During a cycling task, increased out-of-plane joint motion was observed in all children with CP in comparison to their TD peers. Increased out-of-plane motion was earlier observed in 4-year old TD children (Chapter 2). As TD children age, there is a reduction in the amount of out-of-plane motion. However, it seems that as children with CP age, they continue to cycle with increased motion outside the sagittal plane, as adolescents with CP continue to cycle with significant out-of-plane motion (Johnston et al., 2007).

Because cycling requires mainly motion in the sagittal plane, there was little movement seen in the frontal and transverse plane in TD children of 6 years of age and older (Chapter 2). Indeed, the out-of-plane motion is only observed in TD adults when they are fatigued (Dingwell, Joubert, Diefenthaeler, & Trinity, 2008). However, even though the legs may not be moving outside the sagittal plane, forces are applied to the pedal in the direction outside this plane of motion. During the downstroke, experienced adult cyclists produce an external force on the pedals while a small internal force is produced during the upstroke (Ruby, Hull, & Hawkins, 1992). Similar trends in shear forces were observed for the TD children in our study. As a result, the knee ab/adduction and knee rotation torques are usually increased during the same timings (Ruby et al., 1992). Little is known about the hip ab/adduction and rotation, and pronation/supination torques during TD cycling.

The children with CP observed in this current study had increased outward shear forces on the pedals and joint torques in all planes of motion in comparison to their TD peers. Increased joint torques in out-of-plane motion may indicate an increase in joint stress in atypical directions, which can lead to secondary injuries (Daffner, 1978; Piszczatowski, 2008). However, the joint torques produced by children with CP did not exceed 10 Nm, when cycling at a low resistance (5 Watt). The peak joint torques produced in this study were considerably lower than peak torques produced by children with CP in the sagittal plane during walking (Novacheck & Gage, 2007) and running (Davids, Bagley, & Bryan, 1998). Furthermore, the torques produced by the children with CP were also lower than the knee torques produced during walking in the frontal and transverse planes (Stebbins, Harrington, Thompson, Zavatsky, & Theologis, 2010). This indicates that cycling at low resistance leads to less joint stress than walking and running in children with CP, which lowers the chances of stress injuries due to joint loading.

However, when cycling resistance increases, the joint torques are likely to be increased because increased resistance is linked to an increase in workload. Adult cyclists have a workload of 0.3 times their body weight while cycling without resistance, however, workload increases to twice their body weight while cycling against a resistance of 240 W (Ericson & Nisell, 1986). Further research is needed to investigate the effect of an increase in cycling resistance on the joint torques in children with CP. This will help us to understand, whether, at higher resistance, the joint torques during cycling are still less than those associated with other activities i.e. walking.

Adaptability to changes in cadence

Cadence was altered to study the adaptability of children with CP to changes in task demands. Changes in cadences can accentuate biomechanical differences between various populations—for example, between developmental age groups (Korff & Jensen, 2007). In this study, increasing speed was found to profoundly impact jerk cost. Six of the seven children with CP had increased jerk cost in comparison to their TD peers. These data showed that young children with CP had less fluent cycling patterns, as was described previously in older children with CP (Kaplan, 1995). Kaplan (1995) stated that older children with diplegic CP spent more time around BDC compared to the rest of the crank cycle, producing less fluent cycling motion. When the cadence target was increased, the children with CP cycled less fluently than their TD peers.

Jerk cost is a kinematic measurement that denotes physiological demand because increased heart rate correlated with an increase in jerk cost (Fujii & Nagasaki, 1995b). The large increase in jerk cost for children with CP suggests that changes in movement speed leads to higher physiological demand than for their TD peers. Children with CP were unable to cycle at high cadences, thus, we can assume that their less fluent cycling motion was because they reached their upper physiological limits at lower cadences. As a result of the increase in physiological demand, it can be assumed that young children with CP may be unable to cycle for extended periods of time.

Even though there was substantial variability between the different trials, an increased adaptation trend could be observed with an increase in cadence. The children with di-/quadriplegic CP cycled across a larger range of cadences than the children with hemiplegia, even though they had higher involvement (Table 3.1). The increased range of cadences is probably the reason why cadence effects were observed in the joint kinematics and kinetics in children with di- and quadriplegia and not in the children with hemiplegia.

Type of cerebral palsy had an effect on the cycling biomechanics

In this study, we investigated the biomechanics of children with hemi-, di- and quadriplegic CP. A clear distinction was found between the children with hemiplegic CP and the children with di-/quadriplegic CP. The children with di- and quadriplegia cycled at a larger range of cadences with more variability than the children with hemiplegic CP. Children with hemiplegia distinguished themselves by the increase in bilateral asymmetry in work production. Four out of the five children with hemiplegia depended more on their less affected leg than on their more affected leg, indicating that the effects of cycling therapy might be different for children with hemiplegic CP than for children with di-/quadriplegic CP.

To compensate for their more affected leg, children with hemiplegic CP produced more work with their less affected leg. In a related fashion, the joint torques on the less affected side were more elevated than on the more affected side. In clients who have suffered from a stroke, the asymmetry between the affected and non-affected legs during cycling has been observed before (Brown, Kautz, & Dairaghi, 1997; Fujiwara, Liu, & Chino, 2003; Kautz & Brown, 1998). The non-affected leg compensates for this negative work by applying more force to the pedal.

Compensation by the less affected leg has also been observed during walking in individuals with CP, with the less affected leg generating more power in the hip due to spasticity in the more affected leg (Riad, Haglund-Akerlind, & Miller, 2008). However, walking is an open skill and, therefore, pushing harder with the unaffected leg would not mitigate the adverse effects of the other leg. In cycling, compensation in power production is possible due to the characteristics of a cycling task. Pushing hard on the pedal will provide more momentum to pass BDC and help maintain speed during the upstroke and to compensate for the affected leg providing less power to the pedal.

As a result of the increased force on the pedal due to compensation, children with hemiplegic CP pushed outwards on the pedal with their less affected leg, increasing the shear force. Even though during the downstroke it is common to have an outward shear force (Ruby et al., 1992), this shear force was out of proportion in children with CP. This would suggest that the less affected leg not only compensates but also generates excessive force. The compensation for power production in the less affected leg indicates that children with hemiplegic CP depend more on the muscle activity in this leg. As a result, it is possible that they do not strengthen their muscles in their more affected leg as would be expected in a therapeutic cycling session. Depending on the goal of the therapy, one-legged cycling might be preferred for strength training of the more affected leg. To support the child with CP, with the ability to push the pedal around, assistance on the other pedal might be needed.

Future research

Even though young children with CP can cycle at different cadences, their performance window is very small. Future research should focus on the effect of spasticity reduction on the biomechanics of a locomotor skill in children with CP, to create a larger window of cadences in which children can be trained. Spasticity reduction would reduce physical demand and improve adaptability to changes in task demand. As a result, the emphasis of cycling therapy could be focused on training to improve the overall fitness levels of children with CP. Different treatments, such as botulinum toxin (BTX) injections, selective dorsal rhizotomy or oral pharmacological interventions, are used in children with CP to reduce spasticity (Koman, Smith, & Shilt, 2004). BTX injections, in particular, are used as a supplement to physical therapy to improve functional motor skills (Cosgrove, Corry, & Graham, 1994; Koman et al., 2004).

Summary

Children with CP were able to cycle on an ergometer with trunk support, indicating that despite their disabilities, they were able to produce task-specific forces. Many different kinematic and kinetic solutions to a similar cycling task were found among children with spastic CP. However, when task demands (i.e., cadence) changed, the children lacked the adaptability to perform task-specific adjustments to continue cycling successfully. Children with different types and involvements of CP were included in this study, and they all showed differences in cycling biomechanics, indicating that CP classification influenced the results of our study. Due to the different solutions to the cycling task, the fluency of cycling was much reduced. Children with CP had more problems matching muscular with non-muscular forces, especially when cadence increased.

Physical therapists should be aware that cycling on an ergometer is possible for clients with CP, who need to stay active. Cycling is an excellent alternative to walking and running in children with spastic CP, especially because joint stress is low while cycling at low resistance and cadence. However, cycling is significantly more challenging for children with CP than for TD children, and cycling therapy should continue to be applied with careful consideration, depending on the goal of the therapy.

Chapter 4: Study 3

The impact of botulinum toxin treatment on stationary cycling – Two pediatric case studies

INTRODUCTION

More than 85 percent of children who suffer from cerebral palsy (CP) have spasticity (Johnson, 2002). Spasticity is an abnormally increased level of muscle activity during rest and increased resistance to passive movement. With spasticity, the resistance against passive movement increases with velocity and the direction of the joint motion (Sanger et al., 2007). Increased levels of muscle activity lead to atypical motion during the performance of fine and gross motor skills (Himmelmann, Beckung, & Hagberg, 2006). Spasticity can be reduced with medical interventions such as botulinum toxin (BTX) injections, selective dorsal rhizotomy and/or oral pharmacological agents (Koman, Smith, & Shilt, 2004). BTX is a popular treatment for spasticity in children with CP because it can target specific muscles and has reversible effects (Koman et al., 2004; Love et al., 2010). BTX causes neuromuscular blockades that prevent the nerve from releasing acetylcholine to the muscle, with the side effect of reduced muscle tone (Koman et al., 2004). As a result, the muscle is prevented from contracting (Arnon et al., 2001). On average, this effect lasts between 3 and 6 months (Pavone et al., 2016). The determination of which spastic muscle to target with BTX depends on the goal of the treatment (Love et al., 2010). For example, if the goal is to reduce toe-walking the gastrocnemius would be injected with BTX.

BTX is an adjunctive intervention that improves range of motion (ROM) and reduces spasticity creating therapeutic opportunities (Jankovic, 2004; Koman et al., 2004; Love et al., 2010). BTX therapy is used in combination with other treatments like physical therapy to improve functional motor skills, (Graham et al., 2000), potentially strengthen the antagonist muscles (Love et al., 2010) and improve the balance between agonists and antagonists (Edgar, 2001; Koman, Mooney, & Smith, 2000). In children with CP positive effects of BTX on the biomechanics of gait (Sutherland, Kaufman, Wyatt, & Chambers, 1999; Zurcher, Molenaers, & Desloovere, 2001) and upper extremity movements (Bensmail, Robertson, Fermanian, & Roby-Brami, 2010; Mackey, Miller, Walt, Waugh, & Stott, 2008) were observed. No locomotor skill, other than gait, has been studied.

Cycling is frequently used as a therapeutic intervention in children with CP due to the reduced demands on balance control (Fowler et al., 2010) and the opportunity to increase exercise modalities (Mutton et al., 1997). Resistance and speed are manipulated during cycling therapy to improve muscle strength and functional skills (Chen et al., 2012; Fowler et al., 2010; Williams & Pountney, 2007). Young children with CP find it difficult to cycle at high cadences or against a high resistance, so when children with CP can cycle, they cycle with a different technique (Chapter 3). In a previous study, we found that young children with CP cycled with increased plantar flexion, increased out-of-plane motion, increased pedal forces, increased jerk cost (a measure of movement fluency) and higher joint kinetics in all three planes of motion compared to typically developing (TD) peers (Chapter 3). Increasing bilateral asymmetry in joint torques occurred as they relied more upon the least affected leg to produce more work. Children with CP were less adaptable to changes in cadence due to the significant increase in jerk cost at high cadence rates (Chapter 3). It is difficult for the children with CP to adapt to changes in task demand that are used during cycling therapy as their cycling biomechanics differ from those of the TD peers.

The purpose of this study was to investigate whether BTX injections lead to a normalization of cycling for young children with hemiplegic CP. The normalization of cycling has the potential to improve the efficacy of cycling as a training modality for the major muscle groups responsible for locomotor skills. BTX has been shown to be effective in normalizing gait (Boyd, Pliatsios, Starr, Wolfe, & Graham, 2000; Corry, Cosgrove, & Duffy, 1998; Sutherland et al., 1999). The variability in subject-specific characteristics of cerebral palsy makes it difficult to aggregate data across children with CP. Thus, in this study, we provide two case studies of the effects of BTX treatment on the lower extremity kinematics and kinetics of stationary cycling for two children with CP.

The study investigated whether BTX changed the kinematics and kinetics of motion for the leg treated with BTX, (Tx) and examined whether the changes induced by BTX led to a normalization of the performance characteristics to more closely approximate the cycling performance of TD peers, and thus create optimal cycling performances. By changing cadence, we also investigated the effect of BTX treatment on the adaptability to changes in task demands (cadence).

METHODS

Participants

Participants included two 5-year-old children with right hemiplegic CP. Data on these children were collected prior to the BTX treatment and 3 weeks post treatment. To provide a performance reference, the two children with CP were compared to seven TD children. Data on these seven children were presented in Chapter 3. TD participants were recruited to match the CP participants on age (within 2 months of age during the first session), gender, height, weight, and cycling experience. Pre BTX treatment data on these two participants with CP were reported in Chapter 3.

During the first session, the parents of the participants signed an informed consent that was approved by The University of Texas Institutional Review Board and the Institutional Review Board of Dell Children's Medical Center of Central Texas at Austin. Parents completed a questionnaire regarding the child's motor skills, the severity of impairment and their classification on the Gross Motor Function Classification Scale (GMFCS) (Alshryda & Wright, 2014) (Table 4.1). The parents provided the BTX treatment information regarding injection site and dosage as provided by the child's physician (Table 4.2).

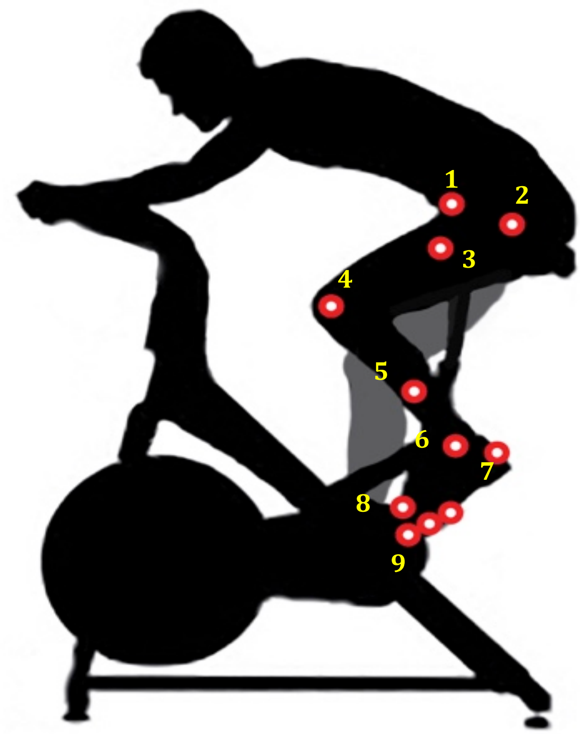
Anthropometric data were obtained during the first session (height, weight, and leg length) and was used to match a child with CP to a TD peer and to adjust the ergometer to a standardized position for each participant (Chapter 3). Spasticity was measured using the Modified Ashworth Scale (MAS) (Bohannon & Smith, 1987) in the treatment leg (Table 4.2).

Equipment and preparation

The participants cycled on a Lode Corival Pediatric cycle ergometer (Lode Corival Pediatric, Lode, Groningen, Netherlands) with resistance set at 5 W. The pedals of the ergometer were customized with cleats and two force transducers in each pedal (model 9251AQ01, Kistler, Winterthur, Switzerland) to collect forces applied to the pedal during cycling. The reaction forces measured at the pedal were collected at 1200 Hz. On each pedal, there were three infrared reflective markers aligned to track the position of the pedals.

Before the participants were seated on the ergometer, 16 reflective markers were attached to the participants' lower legs, eight markers on each side of the body, and three markers were aligned on each pedal (Figure 4.1). A 10-camera passive marker tracking system (Vicon Nexus 1.5.1) was used to collect marker positions during cycling at 120 Hz.

After completion of the first cycling session, children with CP underwent BTX treatment as part of their regular treatment. Participant 1 (CP 1) received BTX injections in the peroneus, medial hamstring (HS) and gastrocnemius (GAS) of her Tx leg. Participant 2 (CP 2) received BTX in the hip adductors, HS and GAS of the right leg (Table 4.2). Three weeks after BTX treatment, CP1 and CP2 attended their second cycling session. The cycling paradigm was constant across both data collection sessions. The children did not cycle at home or during therapy between sessions 1 and 2.



MARKERS LOCATIONS

1. Anterior superior iliac spine
2. Greater trochanter
3. Upper third of the thigh
4. Lateral femoral condyle
5. Lower third of the tibia
6. Lateral malleolus
7. Calcaneus
8. Head of 2^{nd} metatarsal
9. Aligned pedal markers

Figure 4.1: Position of reflective markers during data collection. The locations of the markers on the left side of the participant's body are shown in this figure. The same marker locations were used on the right side.

Table 4.1: Participant information

PARTICIPANT	GENDER	AGE	TYPE SPASTIC CP	GMFCS	HEIGHT (M)	MASS (KG)	RPM	NO. REVOLUTIONS ANALYZED	DAYS BEFORE BTX	DAYS AFTER BTX
CP 1	F	5yr 2m	Right Hemi	I	1.09	17.7	30 40 50	3 5 4	14	21
CP 2	F	5yr 8m	Right Hemi	I	1.13	15.9	60 70	5 5	20	21

Note: GMFCS: Gross Motor Function Classification Scale, RPM: revolutions per minute

Table 4.2: BTX injections for each participant and the scores on the MAS pre and post BTX.

Participant	BTX injection type	Amount of BTX per muscle group	MAS pre BTX	MAS post BTX
CP 1	BTX Type A concentration 100 Units/ml	10 units right peroneus 10 units right HS 40 units right GAS	HS: 1 RF: 0 GAS: 3 TA: 1	HS: 0 RF: 0 GAS: 1 TA: 0
CP 2	BTX Type A concentration 100 Units/ml	20 units right hip Adductors 20 units right HS 40 units right GAS	Hip adduction: 1 HS: 1+ RF: 0 GAS: 2 TA: 0	Hip adduction: 0 HS: 1 RF: 0 GAS: 1 TA: 0

Note: BTX: Botulinum Toxin, MAS: Modified Ashworth Scale, HS: medial hamstring, RF: rectus femoris, GAS: gastrocnemius, TA: tibialis anterior

Data collection and analysis

During the first session, the participants were asked to cycle at a steady state of 30, 40, 50, 60, 70 and 80 revolutions per minute (RPM). The participants were instructed to remain within a range of ± 4 RPM around the target cadence, and data (marker locations and pedal forces) were collected for 25 s once the participant cycled at a steady state. A trial was included for further analysis if the participant produced a minimum of three consecutive revolutions within the target cadence. In Table 4.1 the successful target cadences per participant are shown. During the post BTX session, these successful cadence targets were repeated. In both sessions the participants practiced cycling at a steady-state pace for 5 min without any resistance before data collection; this practice simultaneously served as a warm-up.

Data were analyzed using Matlab (7.0.1) and OpenSim (3.0). Pedal revolutions were selected for analysis if they met the criteria of pace (target + 4 RPM). One of a series of five consecutive revolutions within the specified cadence, or when the subject failed to produce a sequence of five successful revolutions, four or three revolutions were included for further analysis (Table 4.1). The number of revolutions included for further analysis during the post BTX session was matched to the successful number of revolutions during the pre BTX session. A revolution was defined from top dead center (TDC) of the right foot (0° crank angle) until the right foot reached TDC again (360° crank angle).

Jerk cost, a measurement of the fluency of motion (Chang, Wu, Wu, & Su, 2005), was calculated in Matlab, using equation 4.1:

$$Jerk\ Cost = {}^{1}/_{2} \int_{0}^{T} J(t)^{2} dt \qquad \text{(Eq. 4.1)}$$

where J(t) is the jerk, the third derivative of the crank angle and T the movement time of the revolution.

The tangential forces and crank torques were calculated in Matlab to determine the amount of work produced by each leg. A percentage was calculated to observe the proportion of work that was delivered to the pedal by the Tx and the non-treated (non-Tx). Force pedal data were also used to determine the joint power in the sagittal plane produced by each joint of the lower legs using inverse dynamics techniques (Korff, Hunter, & Martin, 2009; Korff, Newstead, van Zandwijk, & Jensen, 2014). The power produced by the muscles around each joint in each is expressed relative to the overall pedal power produced by that leg.

Three-dimensional joint angles and torques were determined using a musculoskeletal model in OpenSim (Delp, Anderson, & Arnold, 2007). The ankle joint angle in the sagittal plane was used to identify phase-plane dynamics in the ankle. The joint torques at the left and the right side of the participants were plotted regarding the crank revolution. The conventions of the crank cycle are defined as 0° and 360° crank angle when the right foot was at top dead center (TDC) and 180° is designated as bottom dead center (BDC).

The data from the TD children were averaged for each cadence. A 95% confidence interval (CI) was calculated around the mean to provide a reference for the atypical cycling characteristics of the two children with cerebral palsy.

RESULTS

Biomechanical analysis TD peers

Cycling consistency TD peers

During the 25 s data collection the TD children were able to cycle at a steady-state for at least five consecutive revolutions at all cadences (Table 4.3).

Table 4.3: Number of consecutive revolutions within the target during the 25 s data collection trial.

Participant	Target cadence (RPM)	No. of consecutive revolutions pre BTX	No. of consecutive revolutions post BTX	% of consecutive revolutions improved between pre and post	No. of consecutive revolutions TD peers (±SD)	Pre BTX no. of consecutive values within 95% CI	Post BTX no. of consecutive values within 95% CI
CP 1	30	3	7	133	11.7 (2.4)	No	No
	40	7	9	29	9.4 (4.3)	Yes	Yes
	50	4	7	75	12.6 (6.6)	No	Yes
CP 2	60	7	11	57	15.0 (2.6)	No	No
	70	9	13	44	21.7 (9.9)	No	Yes

Ankle phase-plane dynamics TD peers

All the phase plane plots for the TD peers had an elliptical pattern, which indicated that the trajectory followed a circular path throughout the cycling motion. A major axis can be distinguished in the elliptical curve, in this case the angular displacement axis, and a minor axis, in this case the angular velocity axis. The phase plane plot for hip velocity versus angle indicated that at TDC, the children transfer from hip flexion to extension, and the opposite occurred at BDC (Figure 4.2A). The major axis in the hip phase-plane plot ranged on average from 39 ° to 85 ° and the minor axis from -135 deg/s to 111 deg/s. The children started to extend the knee before TDC, hence, the positive velocity of the knee at TDC (Figure 4.2B). The children also started to flex the knee before BDC. In the knee phase-plane, the major axis ranged on average from -120 ° to -46 ° and the minor axis from -184 deg/s to 189 deg/s. The children started to plantar flex after TDC (at TDC the ankle velocity was still positive), and they started to dorsiflex the ankle after BDC (at BDC the ankle velocity was still negative, Figure 4.2C). In the ankle phase-plane, the major axis ranged on average from 1 ° to 22 ° and the minor axis from -54 deg/s to 57 deg/s.

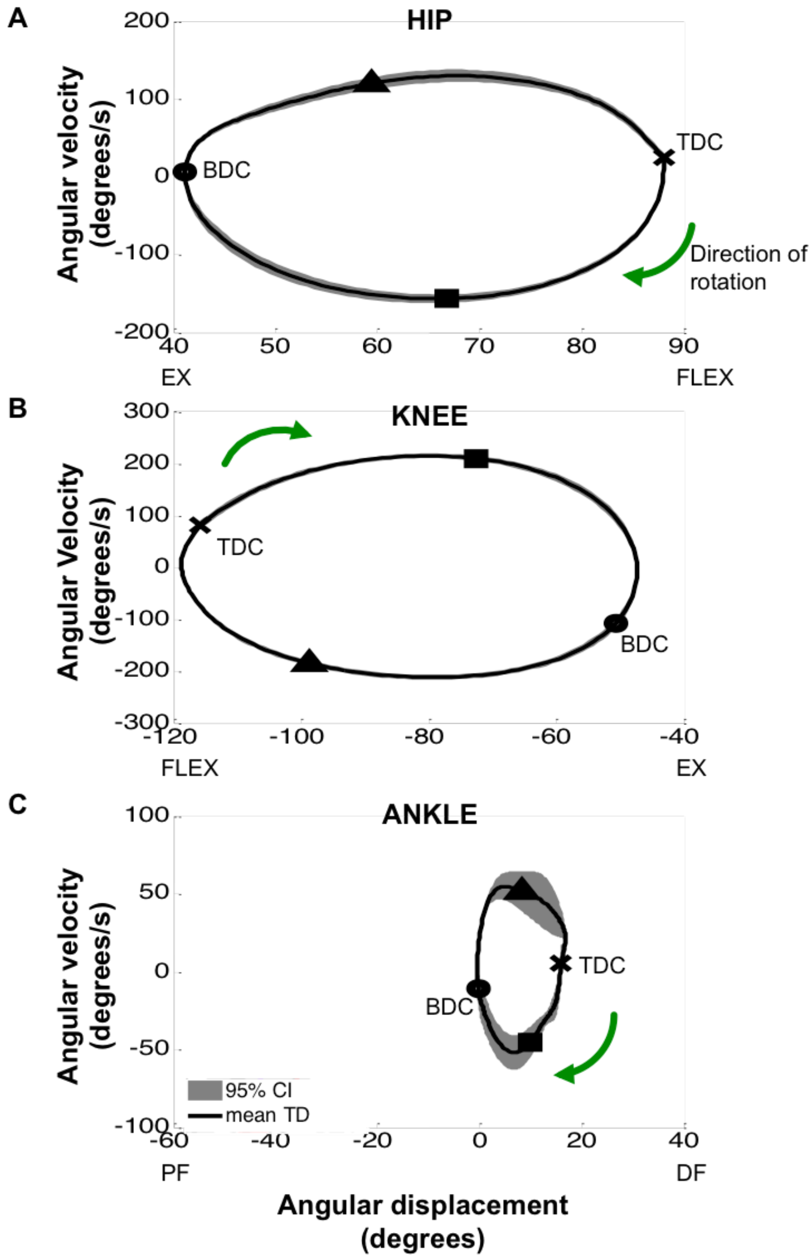


Figure 4.2 Phase dynamics plots for TD peers. Phase-plane dynamics plots for TD peers at 60 RPM at the hip (A), the knee (B), and ankle (C) in the Tx leg before and after BTX treatment and the TD peers. Crosses represent TDC, squares mark the position of 90° in the crank cycle, circles mark BDC and triangles representing 270° in the crank cycle. The direction of rotation for each graph is clock-wards.

Jerk cost TD peers

The jerk cost for the TD peers ranged from 0.5 $deg^2/s^5 \times 10^9$ at 30 RPM to 1.5 $deg^2/s^5 \times 10^9$ at 70 RPM. The upper boundary of the 95% CI stayed below 1.2 $deg^2/s^5 \times 10^9$ during the lowest four cadences and increased to 2.1 $deg^2/s^5 \times 10^9$ when the children reached 70 RPM.

Percentage work done TD peers

The TD children had bilateral asymmetry in the amount of work produced by each leg. They cycled with the highest asymmetry at 30 RPM (lower boundary of 22% and upper boundary of 78%) and with the smallest amount of asymmetry at 70 RPM (lower boundary of 41% and upper boundary of 59%, Table 4.4).

Table 4.4: Percentage work that was done by the Tx leg in comparison to the non-Tx leg for CP 1 and CP 2.

Participant	Cadence (RPM)	Percentage work done pre BTX	Percentage work done post BTX	Percentage work done 95% CI
CP1	30	48	43	22-78
	40	51	42	38-62
	50	54	42	38-62
CP2	60	18	19	36-64
	70	22	289	41-59

Note: The percentage work done by the TD peers was calculated for their dominant leg and then it was mirrored (i.e. flipped around the 50% mark) to find the lower boundary on the non-dominant side to calculate the 95% CI.

Joint torques TD peers

During the cycling revolution, the TD children produced a hip flexion and plantar flexion torque at TDC (Figure 4.3A). Throughout the downstroke, the power production phase, the children produced a peak knee flexion torque around 90° crank angle (on average a knee extension torque of 4 Nm) and a plantar flexion torque (on average 4 Nm), to push the pedal downwards. Towards BDC, the muscles around the hip were creating an extension torque, which still existed at BDC. This hip extension torque is created to push the pedal downwards during the power stroke and to push the pedal backward to pass BDC (Zajac, Neptune, & Kautz, 2002). The highest hip extension torques could be observed just before BDC (with an average peak value of the upper boundary of the 95% CI of 4 Nm). During the upstroke, knee flexion and ankle plantar flexion torques were produced.

Small torques were created in the transverse and frontal plane including: hip and knee abduction as well as adduction torques (smaller than 4 Nm), internal as well as external torques (smaller than 2 Nm), and subtalar pronation as well as supination torques (smaller than 2 Nm).

Joint powers TD peers

TD children produced the most power during the extension phase (downstroke) of the crank cycle (on average 91% of total produced pedal power, Figure 4.3). During the downstroke the muscles around the hip produced 82% of the net overall pedal power, the muscles around the knee produced 3% and the muscles around the ankle 16%. During the upstroke (flexion phase) power was absorbed from the system by the muscles around the hips (on average 15% absorbed from overall produced power), while the knees and ankles produce additional power (on average 21% and 3% of the overall net power, respectively) to help the opposite leg by pulling the pedal up.

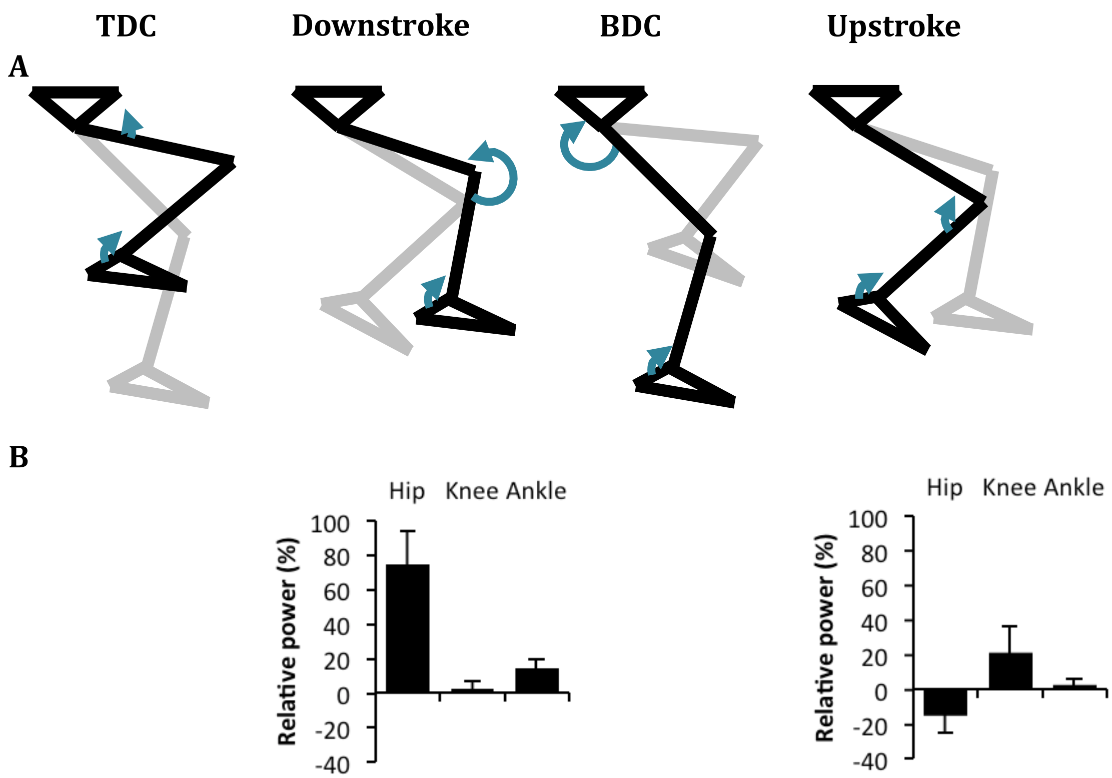


Figure 4.3: Joint kinetics produced by TD peers. Joint torques are presented at TDC, 90° crank cycle, BDC, and 270° crank cycle (A). Hip, knee, and ankle torques are displayed as blue circular arrows, which indicate the direction of the created torque. Joint powers during the down- and upstroke are presented in the bar graphs (B). The data displayed were the averages over all performed cadences.

Biomechanical analysis CP 1

Cycling consistency CP 1

Pre BTX, CP 1 was unable to cycle successfully for five consecutive revolutions at 30 and 50 RPM (Table 4.3). At these cadences, CP 1 produced a lower number of consecutive revolutions than the TD peer. After treatment, CP 1 was able to cycle at least seven consecutive revolutions at a steady-state. At this time, only the number of successful consecutive revolutions at 30 RPM fell outside the 95% CI.

Ankle phase-plane dynamics CP 1

Similar to the TD peers, the phase-plane plots for the hip and knee of CP 1 pre BTX, had elliptical shapes. While the TD peers started extending their hips after TDC and flexing around BDC, CP 1 started to extend her Tx hip before TDC and started to flex her Tx hip after BDC (Figure 4.4A). Pre BTX, the hip phase-plane trajectory for CP 1 was increased in comparison to the trajectory of the TD peer. The major axis was on average increased by 3 ° and the major axis increased by 33 deg/s. CP 1 started to flex her knee around BDC; this was later than the TD peers since they started to flex the knee before BDC. On average over all cadences, the minor axis of the knee phase-plot was decreased by 13 deg/s, and the major axis was decreased by 9 ° (Figure 4.4B). In the knee, the TD peers reached the maximum values on in phase-plane before CP 1. Hence, the positions markers of CP 1 are located on the phase-plane plot before the markers of the TD peers. The ankle phase-plane showed that pre BTX, CP 1 dorsiflexed her ankle at a constant angular velocity, which led to a less perfect elliptical shape (Figure 4.4C). Pre

BTX, CP 1 also cycled with on average 45° more plantar flexion than the lower boundary of the 95% CI. The major axis of the ankle phase-plane plot of CP 1 was on average 10 ° increased and the minor axis 94 deg/s increased.

Post BTX a change toward approximating TD peers in size of major and minor axes occurred in the hip and ankle (the major axes for both the hip and the ankle reduced to differences with the TD peers of smaller than 1 °; the minor axis reduced to a difference of 12 deg/s and 51 deg/s, respectively). The knee phase-plane plot was also reduced (reduction of 7 ° in the major axis and 32 deg/s in the minor axis in comparison to pre BTX values), unlike the phase-plane plots for the hip and ankle, this reduction led to values further away from the plot size of the TD peers than before BTX. The markings of the specific locations in the crank cycle corresponded to the markings of the TD peers, indicating that post BTX the child with CP had the same phase-plane trajectory as the TD peers. In the Tx ankle no constant angular velocity could be observed post BTX, which led to a more elliptical phase-plane post BTX than before treatment. The reduced size and improved shape led to an ankle phase-plane plot post BTX that was more similar to an elliptical shape, like the phase-plane of the TD peers. Post BTX the increased plantar flexion of the treated leg persisted.

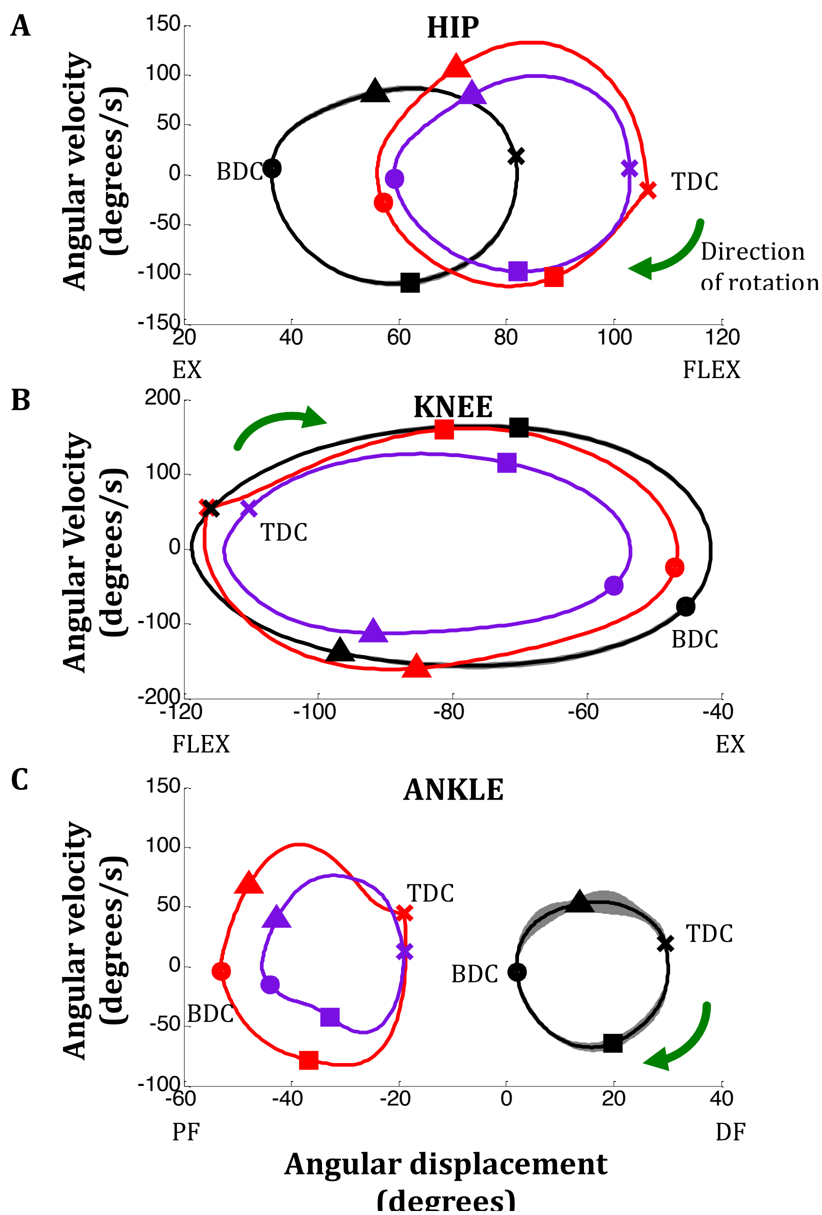


Figure 4.4: Phase dynamics plots for CP 1. Phase-plane dynamics plots for CP 1 at 40 RPM at the hip (A), the knee (B), and ankle (C) in the Tx leg before and after BTX treatment and the TD peers. Crosses represent TDC, squares mark the position of 90° in the crank cycle, circles mark BDC and triangles representing 270° in the crank cycle. The direction of rotation for each graph is clock-wards.

Jerk cost CP 1

Administration of BTX led to a reduction in jerk cost. Pre BTX injections it was observed that CP 1 had higher jerk cost than their TD peers (Figure 4.5). Before the injections CP 1 had problems with cycling fluently, which led to jerk costs (ranging from 2 deg^2/s^5 $x10^9$ at 30 RPM to 8 deg^2/s^5 $x10^9$ at 50 RPM) that varied from 3.6 to 10.0 times larger than the jerk costs produced by the TD peers. The jerk cost was exponentially increased between 30 and 50 RPM (an increase of 271%). Post BTX treatment a substantial reduction in jerk cost was observed; jerk cost was reduced by 29% at 30 RPM and with 88% at 50 RPM. These post treatment values approached values produced by the TD peers (post BTX, jerk cost produced by CP 1 ranged from values that were within the 95% CI or only a modest 2.6 times larger than the upper boundary of the 95% CI).

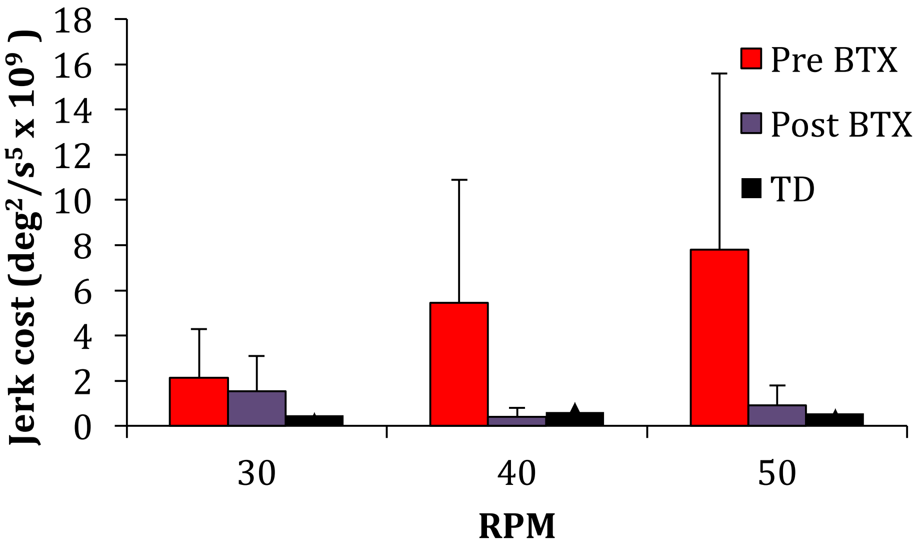


Figure 4.5: Jerk cost for CP 1 before and after BTX treatment. The error bars for the CP 1 (bars with cap) represent within-trial variability, and the error bars for the TD peers (bars with arrow) represent the 95% confidence interval.

Percentage work done CP 1

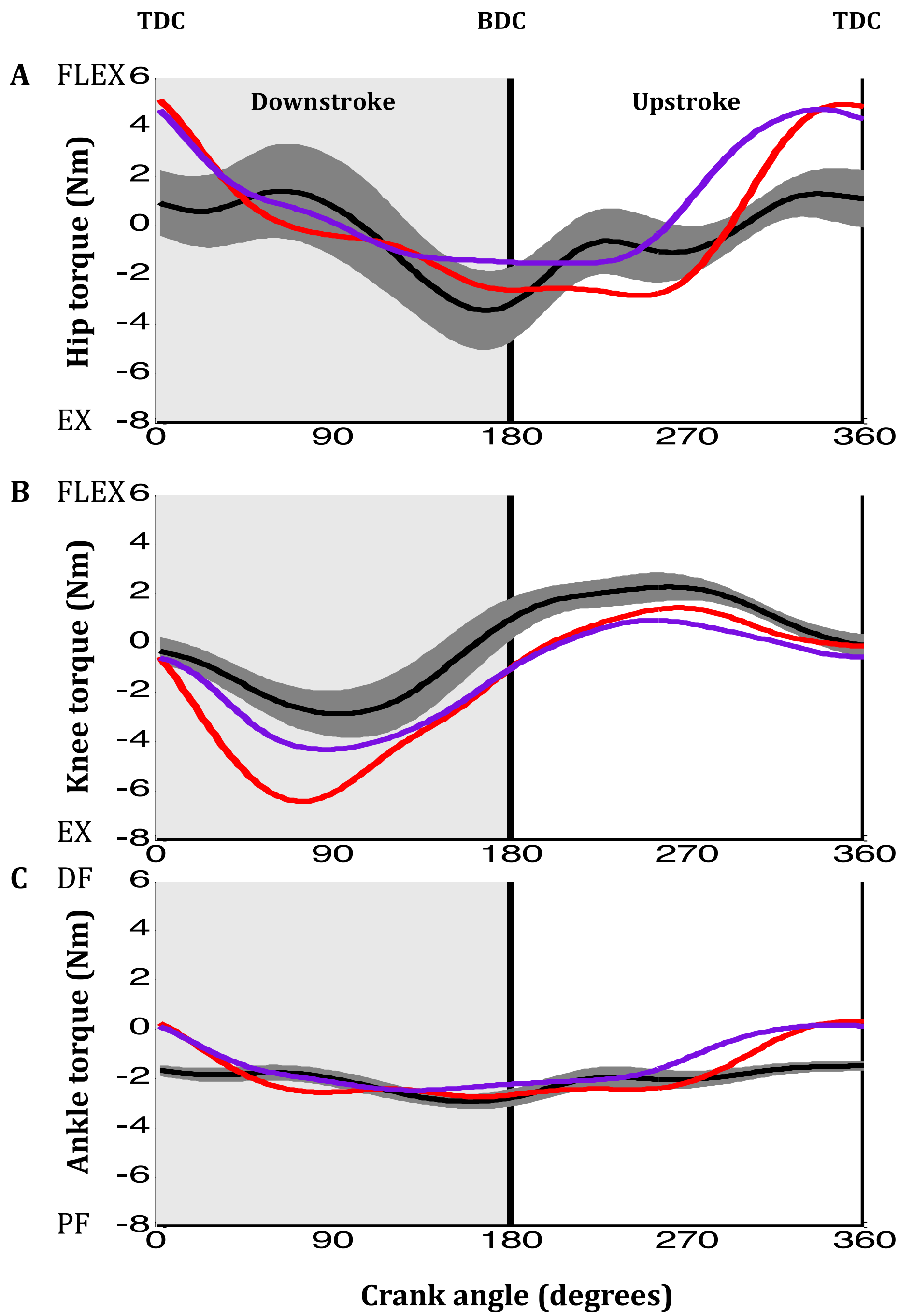
Before BTX treatment, CP 1 produced a similar amount of work with her treated leg as with her non-treated leg (on average 51% work is done by the Tx leg, Table 4.4). The distribution of work between both legs fell within the 95% CI of the TD peers. After BTX treatment, small changes were observed in work generated by each leg, in comparison to pre BTX session. CP 1 produced less work with her treated leg post BTX than before treatment (average shift from 51% to 42% work produced by treated leg). During both pre and post BTX session, CP 1 had asymmetry in work produced by each leg that fell within the 95% CI of the TD peers.

Joint torques CP 1

Increased flexion/extension torques at the hip and knee were observed in CP 1 pre BTX injection (Figure 4.6A-C). The highest peak torque was observed during knee extension in the treated leg (9 Nm, which was 2.3 times larger than upper boundary of 95% CI). For the treated leg of CP 1, the total sum of applied torque during the power stroke was also increased. At all three cadences, this child produced on average 1.9 times more knee torque during the downstroke than the TD peers. At 30 RPM, the hip and ankle torque were increased (respectively, 1.6 and 1.1 times larger than the TD peers). CP 1 had also increased joint torques in all motions in the frontal and transverse plane (Figure 4.6D and 4.6E). Hip and knee adduction at 30 RPM were the highest observed torques in the treated leg (4 Nm and 4 Nm, respectively; both 2.1 times larger than 95% CI).

BTX injections reduced the joint torques at all joints in the treated leg (with reductions up to 4 Nm in the hip, 6 Nm in the knee and 1 Nm in the ankle). The total amount of joint torque during the entire downstroke was reduced for all joints and at all cadences (with a maximum reduction of 85% for hip torque at 30 RPM). The joint torque produced by CP 1 at 30 and 40 RPM were reduced post BTX to within the 95% CI. At 50 RPM the hip and knee flexion torques in the treated leg were reduced in comparison to pre BTX settings, however, they were still increased in comparison to the TD peers (Figure 4.4). The joint torques in the frontal plane were also reduced after treatment for each participant. CP 1 had reduced joint torques at all cadences, with the largest reduction at 30 RPM (reduction in torque up to 80%).

The large reduction in joint torques production led to values in the frontal and transverse plane similar to the TD peers. In CP 1, at 30 and 40 RPM, all the joint torques in the treated leg were reduced to within the 95% CI. At 50 RPM the torques were reduced but not all torques fell within the 95% CI. The largest torque post treatment that fell outside the 95% CI was the hip internal rotation torque (peak torque of 2 Nm).



TDC
BDC
TDC
A
FLEX
EX
Downstroke
Upstroke
Hip torque (Nm)
B
FLEX
EX
Knee torque (Nm)
C
DF
PF
Ankle torque (Nm)
Crank angle (degrees)

Figure 4.6

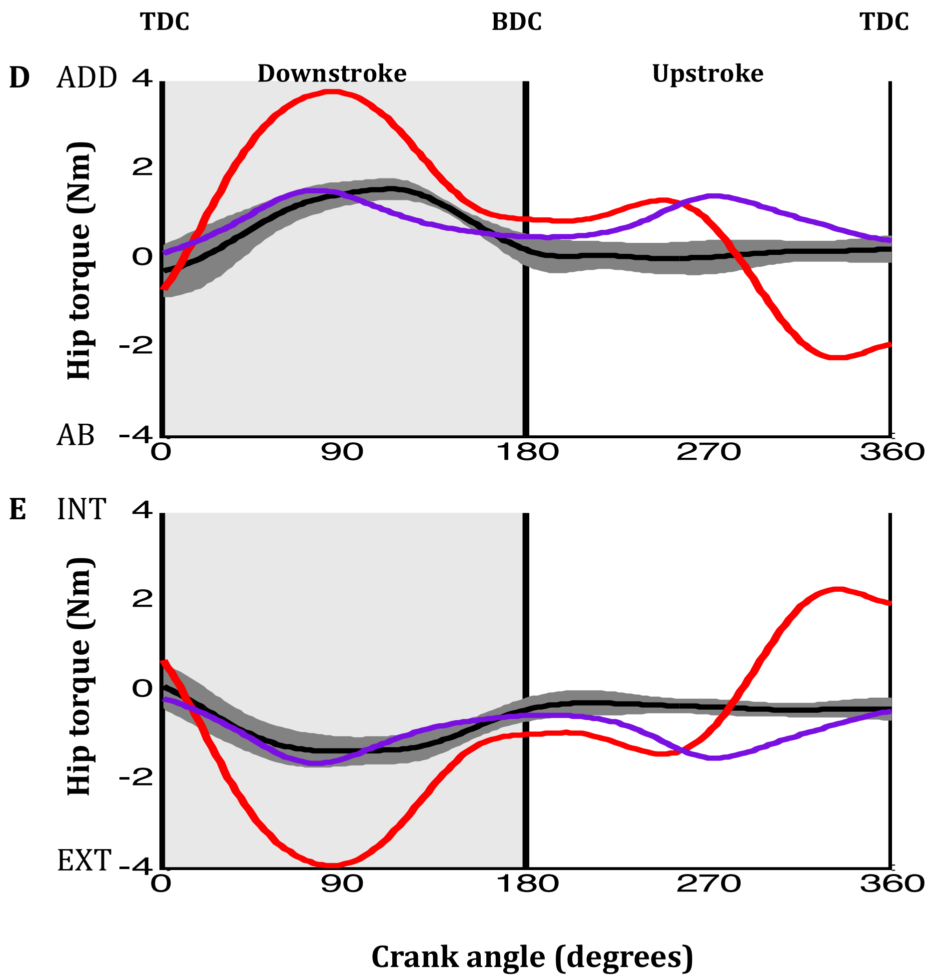


Figure 4.6: Joint torques produced by CP 1. Hip flexion (A), knee flexion (B) and ankle flexion (C) torques produced during cycling at 40 RPM, and hip adduction (D) and hip rotation (E) torques produced during cycling at 30 RPM are shown.

Joint powers CP 1

Before BTX treatment CP 1 produced 80% of the total amount of pedal power with her treated leg during the downstroke (Figure 4.7). The percentage of overall pedal power, during a whole revolution, produced during the downstroke decreased with an increase in cadence (from 101% at 30 RPM to 73% at 50 RPM). CP 1 produced 94% of the total amount of power during hip extension at 30 RPM with her treated leg. This reduced to 77% at 40 and 50 RPM. At the 40 and 50 RPM more knee flexion power was produced (on average 24% in contrast to -8% at 30 RPM).

BTX injections led to an increase in the percentage of pedal power produced during hip extension in the treated leg of CP 1 (on average an increase of 25%) and more absorption of power during knee extension (on average 70% more absorption).

Post treatment, CP 1’s right and left leg power production was closer to the TD peers than before BTX i.e. the participants produced power during the same phase as the TD peers.

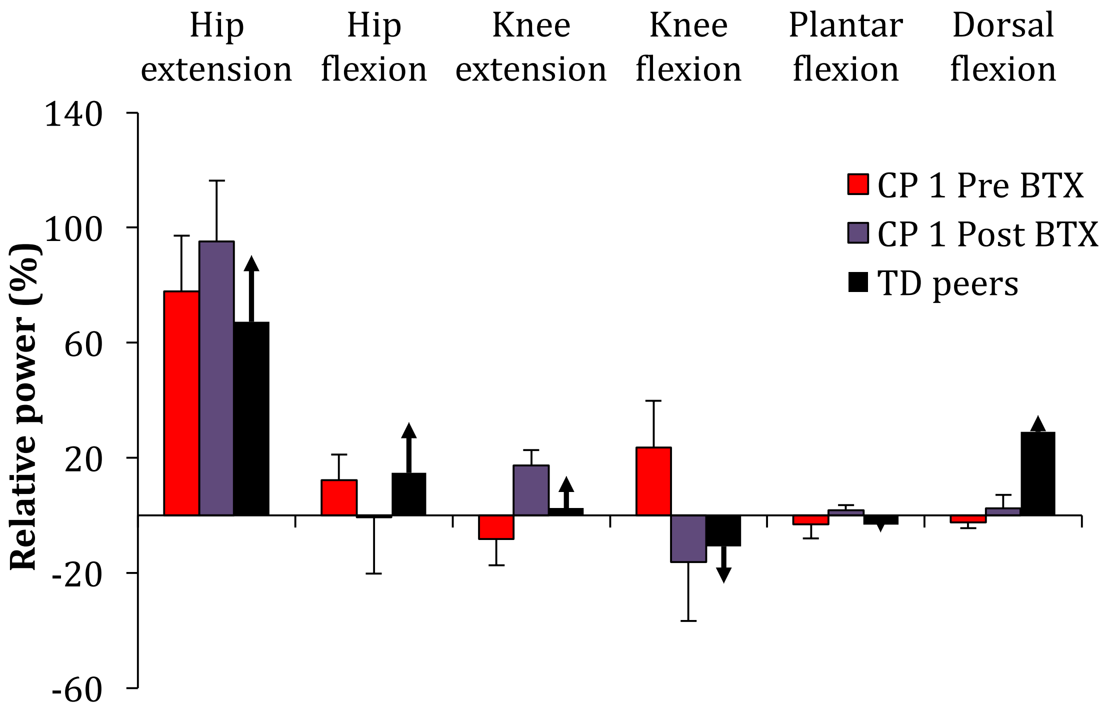


Figure 4.7: Joint powers produced with the treated leg by CP 1 at 40 RPM. The error bars for the CP 1 (bars with cap) represent within-trial variability, and the error bars for the TD peers (bars with arrow) represent the 95% confidence interval.

Biomechanical analysis CP 2

Cycling consistency CP 2

CP 2 cycled more than the desired five consecutive revolutions at target cadences before treatment (at least 7 consecutive revolutions, Table 4.3). Even though CP2 cycled successful for more than 5 revolutions, at both 60 and 70 RPM, the number of successful revolutions was smaller than the number produced by the TD peers. At both cadences in the post BTX session CP 2 cycled longer at the steady state than pre BTX (increased by at least four successful revolutions). The number of revolutions produced post BTX during the 70 RPM trial fell within the 95% CI of the TD peers.

Ankle phase-plane dynamics CP 2

The hip and knee phase-plane plot for CP 2 had an elliptical shape pre BTX (Figure 4.8A and 4.8B). The hip phase-plane plot for CP 2 was larger than the hip phase-plane plot for the TD peers (major axis 2 ° larger and minor axis 15 °/s larger). The knee phase-plane plot for CP 2 was decreased in comparison to the TD peers (major axis 11 ° smaller and minor axis 62 deg/s smaller). CP 2 had similar timings of hip and knee angular position and velocity as compared to the TD peers (the marking of TDC and BDC, etc. are at the same locations on the curve).

Pre BTX, CP 2 alternated between dorsiflexion and plantar flexion in the treated ankle during the downstroke, while TD peers only plantar flexed their ankle during the downstroke. This alternation ensured that the trajectory crossed itself between TDC and BDC (Figure 4.8C). At TDC, CP 2 produced the highest (88 deg/s) angular velocity, indicating that she was dorsiflexing her ankle at the point the TD peers started to plantar flex their ankles. CP 2 cycled pre BTX with increased plantar flexion (on average 33 ° more flexion).

Except a small shift (of 11 deg/s on the minor axis) in knee flexion, the phase-plane plots for the hip and knee were not largely influenced by BTX treatment. The hip plot was increased in the major axis by 4 ° and in the minor axis by 7 deg/s. The knee phase-plane plot was reduced by 2 deg/s at the minor axis. The ankle was affected by the treatment. CP 2 had a reduction in plantar flexion in the treated leg (on average a reduction of 25°). The crossing that was observed in the phase dynamics plot of CP 2 disappeared, indicating that the child was no longer alternating between dorsiflexion and plantar flexion. The reduction in plantar flexion in the injected leg of CP 2 also led to a more typical ankle motion. However, the ankle was still 8° more plantar flexed than the lower boundary of the 95% CI. Furthermore, at TDC, CP 2 was not producing the highest angular velocity as was seen pre BTX. CP 2 still did not start plantar flexion at this point.

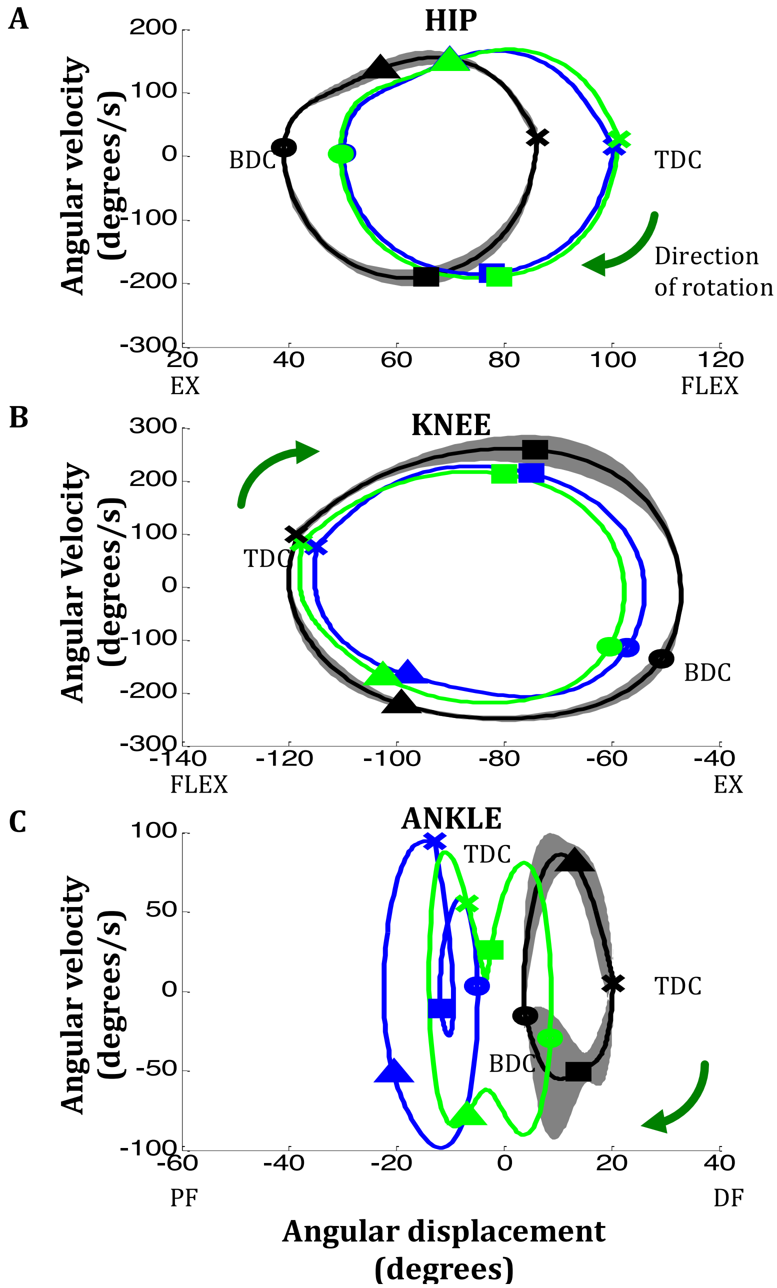


Figure 4.8: Phase dynamics plots for CP 2. Phase-plane dynamics plots for CP 2 at 70 RPM at the hip (A), the knee (B), and ankle (C) in the treated leg before and after BTX treatment and the TD peers. Crosses represent TDC, squares mark the position of 90° in the crank cycle, circles mark BDC and triangles representing 270° in the crank cycle. The direction of rotation for each graph is clock-wards.

Jerk cost CP 2

CP 2 produced pre BTX jerk cost (ranging from 1.1 deg^2/s^5 $x10^9$ at 60 RPM to 7.8 deg^2/s^5 $x10^9$ at 70 RPM) that was between 1.1 and 2.1 times larger than the jerk cost produced by the TD peers (Figure 4.9). This participant experienced an increase in jerk cost as a function of an increase in cadence (an increase of 72%). Post treatment, CP 2 had no reduction in jerk cost at 60 RPM, however, at 70 RPM she there was a reduction. Post BTX there was no observed increase in jerk cost in response to an increase in cadence. The jerk cost values post treatment were 1.1 to 1.2 times larger than the 95% CI of the TD peers.

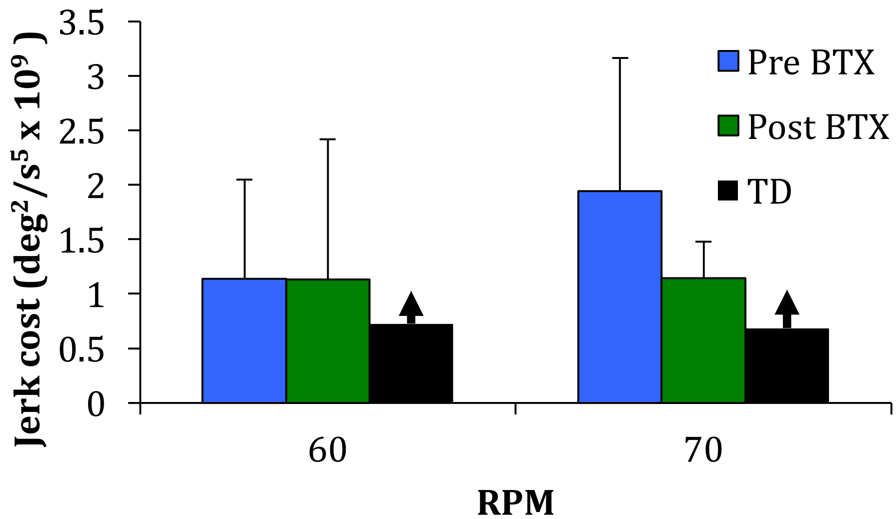


Figure 4.9: Jerk cost for CP 2 before and after BTX treatment. The error bars for the CP 2 (bars with cap) represent within-trial variability, and the error bars for the TD peers (bars with arrow) represent the 95% confidence interval.

Percentage work done CP 2

CP 2 produced only a small amount of the total work done with her treated leg (on average only 20% of the total work done was produced by the treated leg, Table 4.4). CP 2 produced similar work pre and post BTX at 60 RPM, 18% and 19% respectively. At 70 RPM, CP 2 produced more work with the treated leg, in comparison to the non-treated leg, post treatment (22% versus 29%). The increased asymmetry fell outside the 95% CI of the TD peers both pre and post BTX. However, due to the 7.1% less asymmetry post BTX at 70 RPM, the value for asymmetry was 36.0% closer to the 95% CI.

Joint torques CP 2

The highest peak for CP 2 was observed during hip extension (a hip flexion torque of 8.8 Nm, which was 2.1 times larger than the peak value of the TD peers) and knee extension (a knee extension torque of 8.7 Nm, which was 2.4 times larger than the peak value of the TD peers, Figure 4.10A and 4.10B). The overall sum of joint torques during the power phase were increased in comparison to the TD peers; the largest differences between CP 2 and the TD peers were observed at 70 RPM during the extension phase (hip flexion torque on the treated side was increased 6.6 times in comparison to TD peers). In the ankle, CP 2 continuously produced a dorsiflexion torque, which was opposite from the plantar flexion torque produced by the TD peers (Figure 4.10C). In the frontal and transverse plane CP 2 had increased hip and knee adduction and external rotation torques in her treated leg at both cadences (on average 1.6 times larger than TD peers, see Figure 4.10D and 4.10E). At 60 RPM her hip adduction torque was 2.5 times larger than the torque of the TD peers and at 70 RPM her hip rotation was 1.6 times larger.

Post BTX, all the torques created at the joints of the lower extremities were reduced for CP 2. In her treated leg CP 2 had a reduction of the peak flexion torques up to 7.9 Nm. However, in comparison to pre BTX values an increase in extension torque was observed at the ankle at 60 RPM (an increase of 5.7 Nm) and at the hip and knee during cycling at 70 RPM (an increase of 7.9 and 2.8 Nm, respectively). The sum of joint torque during the power phase was reduced for all joints over the whole extension phase with a maximum reduction of 123% for hip torque at 70 RPM, i.e. an opposite torque was created post BTX. The largest reduction in torques in the frontal and transverse plane was observed in knee adduction. CP 2 had a 40% reduction in knee adduction torque at both cadences and a 21.9% decrease in hip adduction torque at 60 RPM. An increase in hip internal rotation and hip and knee abduction was observed (on average 0.9 Nm increase).

In CP 2 the reduced flexion torques approached the 95% CI or were reduced to within the 95% CI. However, at 60 RPM plantar flexion torques in the treated leg were increased post treatment compared to the TD peers (1.2 Nm larger than the TD peers for the treated leg). The same happened for hip and knee extension torques in the treated leg at 70 RPM (2.5 and 0.8 Nm larger in comparison to the TD peers).

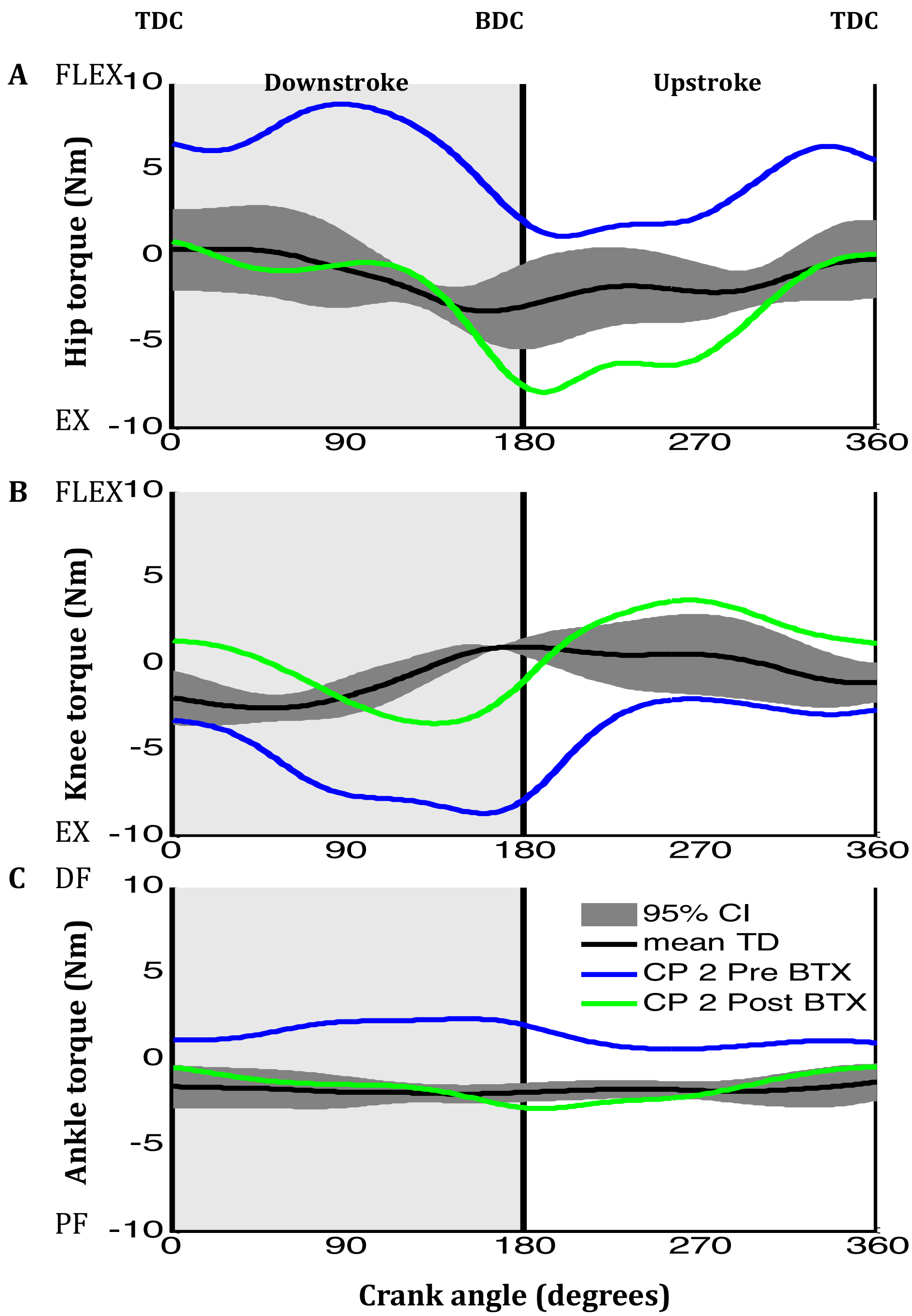

TDC
BDC
TDC
A
FLEX
10
Downstroke
Upstroke
5
0
-5
EX
-10
Hip torque (Nm)
0
90
180
270
360
B
FLEX
10
5
0
-5
EX
-10
Knee torque (Nm)
0
90
180
270
360
C
DF
10
5
0
-5
PF
-10
Ankle torque (Nm)
95% CI
mean TD
CP 2 Pre BTX
CP 2 Post BTX
0
90
180
270
360
Crank angle (degrees)

Figure 4.10

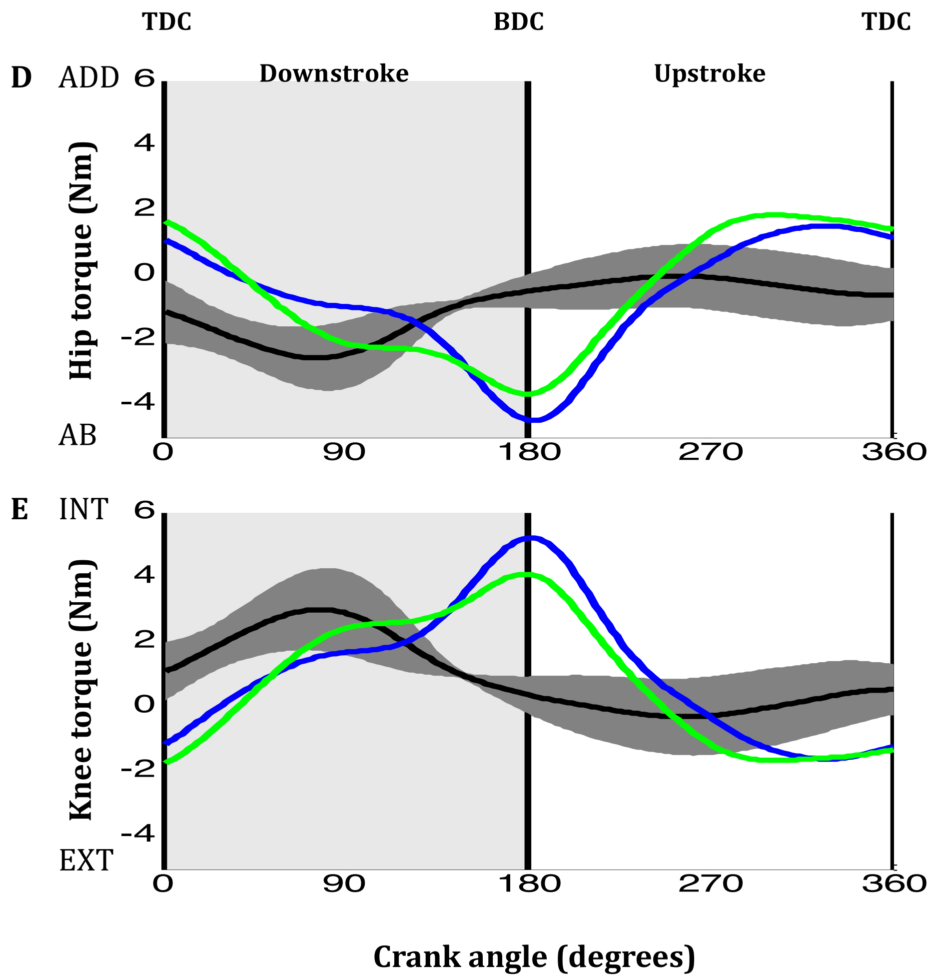


Figure 4.10: Joint torques produced by CP 2. Hip flexion (A), knee flexion (B), ankle flexion (C), hip rotation (D) and knee adduction (E) torques produced during cycling at 70 RPM are shown.

Joint powers CP 2

CP 2 produced powers that were opposite from those produced by the TD peers with the treated leg pre treatment (Figure 4.11). For example, while TD peers produced powers during hip extension, CP 2 absorbed powers during this phase. At 60 RPM, CP 2 absorbed most of her power during the extension phase (-173% of the overall pedal power in the hip and -366% in the knee), which decelerated the pedal, producing most power during the flexion phase (216% in the hip and 429% in the knee) which allowed CP 2 to pull the pedal up. Positive power was produced during plantar flexion at this cadence, while power was absorbed during dorsiflexion (34% and -41%, respectively). At 70 RPM the extremes observed during 60 RPM were reduced.

Post BTX CP 2 produced hip and knee extension power, while pre BTX she was absorbing power. Power was absorbed opposite that of pre BTX power production during the flexion phase. CP 2 produced power during the same phase as the TD peers post treatment.

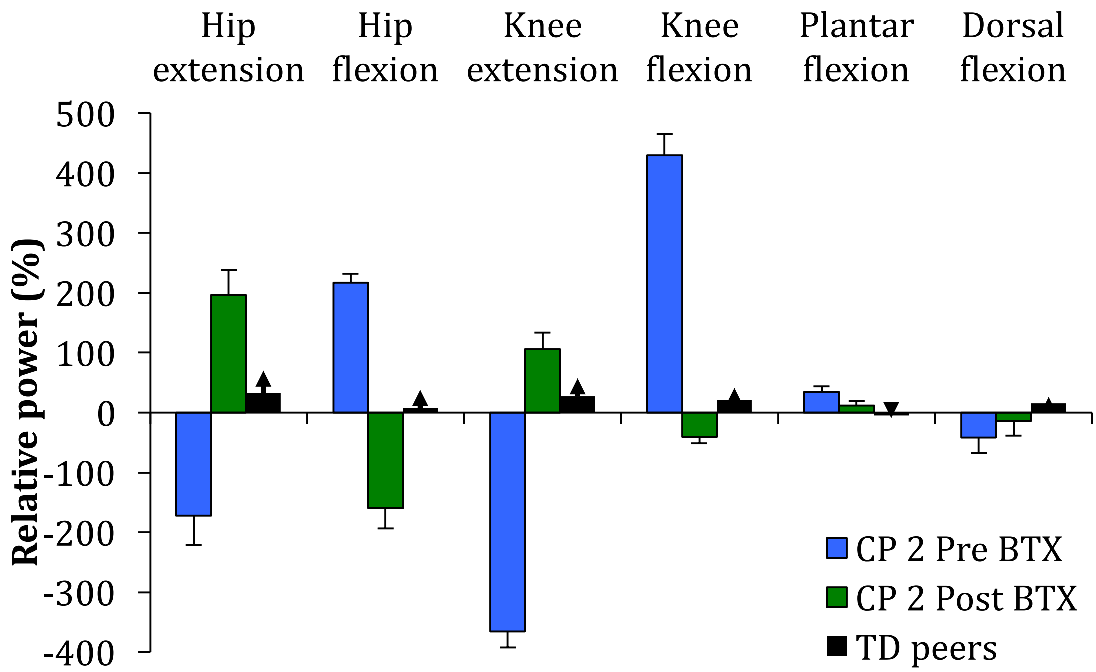


Figure 4.11: Joint powers produced with the treated leg by CP 2 at 60 RPM. The error bars for the CP 1 (bars with cap) represent within-trial variability, and the error bars for the TD peers (bars with arrow) represent the 95% confidence interval.

Summary

Both CP 1 and CP 2 cycled more “typical” post BTX than before treatment. The improved cycling consistency showed that CP 1 and CP 2 found it easier to perform the cycling tasks during the second session. The BTX treatment influenced both children on the ankle phase-plane, but less on the hip and knee phase-planes. Furthermore, post treatment both children cycled with improved fluency, reduced peak and overall joint torques and a more typical joint power production.

DISCUSSION

This study depicts how BTX treatment can support cycling-based training in children with CP. Cycling proved to be a useful motor skill to investigate pre and post BTX treatment as task requirements can be controlled across sessions due to the constraints of the ergometer and the repetitive aspects of cycling. BTX led to improvements in cycling for both children included in this study. BTX also led to greater consistency in meeting cadence demands. Further, BTX led to changes in the kinematics and kinetics of motion, producing more typical cycling characteristics.

The elliptical patterns of the phase-plane plot of the TD peers indicated that smooth accelerations and decelerations occurred with reversals only at the motion extremes during typical cycling (Winstein & Garfinkel, 1989). The deviation away from the elliptical phase-plane in the ankle, pre BTX, showed that there was a reduction in proper timing of the ankle movement during the cycling task. The finding that the ankle trajectory crossed itself for CP 2 is an indication that there is a sudden interruption of forces opposing motion (Winstein & Garfinkel, 1989). Furthermore, certain phase dependent features of normal cycling were absent. For example, CP 2 displayed dorsiflexion in the ankle at TDC, while the TD peers started to plantar flex the ankle. In the knee phase-plane plot of CP 1, the changing directions of movement occurred later than the TD peers displaying a similar lack of features. Post treatment, the elliptical trajectory of the phase plane was smoother for CP 1 and did not cross itself anymore for CP 2. Further, there was a shift towards normal phase dependent features post BTX. The change toward approximating TD peers indicated that the motion control in the ankle was improved due to precisely timed muscle actions.

BTX treatment also had a positive effect on cycling fluency. Both the HS and the GAS of the CP affected right side were treated with BTX, and each had a different function during cycling. In adult cyclists, it was found that the primary role of the GAS is to ensure that the energy generated by the hip and knee muscles (gluteus maximus and vastus medialis) is delivered to the crank (Zajac et al., 2002). This transfer of energy happens during the extension phase of the crank cycle as the plantar flexion action from the GAS is opposing the dorsiflexion motion of the gluteus maximus and vastus medialis. The HS has a significant role during the extension-to-flexion phase (around BDC) of the crank cycle as it maintains a fluent cycle motion by pushing the pedal backward. In young TD children (5-7 years) the power generated during cycling at cadences of 90 RPM or lower is similar to the power generated in adults (Korff & Jensen, 2007). This indicates that in young TD children the muscle functions are likely the same as in adults at low speeds. Because the HS is responsible for fluent motion, it is not surprising that when muscles in the lower extremity suffer from spasticity movement fluency is affected.

In cycling, motion fluency is reduced in most children with hemi-, di- and quadriplegic CP (Kaplan, 1995) (Chapter 3). This loss of fluency is exacerbated as cadence increases. Kaplan showed that older children with diplegic CP spent a disproportionate amount of time around TDC and BDC in comparison to the flexion and extension phase of the crank cycle (Kaplan, 1995). Considering that the HS ensures cycling fluency around BDC by resisting the crank from deceleration (Zajac et al., 2002), it was not unexpected that the cycling fluency improved after spasticity was reduced by BTX. The HS was a targeted muscle for both cases in this study. The BTX-induced reduction in spasticity facilitated the pull through BDC. The measured effect was in the reduction of jerk cost. This improved movement fluency corresponds to previous studies in upper extremity motion in which MAS in patients with CP was positively correlated to jerk cost during reaching tasks (Chang et al., 2005). A similar improvement in reaching was observed in stroke patients (Bensmail et al., 2010). The reduction in jerk cost increased the fluency allowing the children to adjust better to the changes in task demand (i.e., cadence).

The improvement in fluency provides another benefit. Jerk cost has been correlated with energy efficiency. In a bimanual cranking task, decreased motion fluency led to higher heart rate and lower oxygen uptake (Fujii & Nagasaki, 1995; Maruyama, Nakamura, Nagasaki, Ito, & Hashizume, 1987). As motion is more fluent after BTX treatment, we can assume that the children with CP have a reduction in their physiological parameters, leading to less physical demands.

The improved fluency was not due to changes in forces applied by each leg to the crank. The children produced the same percent of total work by the treated leg during the post BTX session. This shows that BTX treatment had little effect on the bilateral asymmetry during cycling. It is important to remember, however, that we selected the "successful" cycling revolutions – the revolutions that met the cadence requirements. Since the study "forced" the child to cycle at a pre-set cadence, it is possible that they favored one leg to generate the rhythm. However, this did not prevent the children from producing more typical joint kinetics. Pre BTX and post, the issue was how the power was generated to move the crank. Changes in both the joint torques and the joint powers demonstrated that BTX facilitated a modification in the production of the crank cycle. Post BTX the joint kinetics more closely approximated the kinetics observed in the cycling of TD.

The net joint torques represents the resultant action of all muscles and other joint structures. For example, the passive resistance of ligaments and tendons and the net joint power is the summed power that is produced by these joint torques (Zajac et al., 2002). Even though these parameters do not provide us with specific information about co-activation, dynamic coupling and energy transfer via biarticular muscles (Zajac et al., 2002) increased joint torques and powers are assumed to be related to muscle effort (Andrews, 1981). In post BTX, the peak joint torques were reduced in comparison to pre BTX and approached values similar to the TD peers. These reductions indicate that the muscle effort during cycling and the stress on the joints were reduced following treatment with BTX. The reduced joint torques were not only observed in the plane of motion but also in the frontal and transverse plane. In addition to BTX injections in the HS and GAS, both participants received BTX treatment to muscles that play an ancillary role in cycling (the hip adductors and peroneus). In contrast to HS and GAS, the role of these muscles relates largely to the out-of-plane motion. Spasticity reduction in these muscles likely contributed to the decreases in torques in the frontal and transverse planes. The result that BTX treatment contributes to a reduction in joint torques is in line with previous studies examining the influence of BTX treatment on joint torques during gait.

These studies showed that during walking, post BTX, the maximum ankle flexion torque before mid-stand was reduced in comparison to pre BTX walking (Boyd et al., 2000; Zurcher et al., 2001). Due to spasticity, children showed a 'double bump' (an extra initial plantar flexion torque peak before the usual mid-stand torque peak) in the ankle plantar flexion torque when spasticity was not treated (Galli, Crivellini, & Santambrogio, 2001). BTX treatment decreased the first bump in the ankle torque in most children with hemiplegic and diplegic CP (Boyd et al., 2000).

The changed timings of the power production, post BTX, indicates that the children in this study developed power like TD children of the same age. Muscle adjustments occur if a locomotive skill cannot be performed optimally and is not entirely the result of spasticity. Experienced cyclists have increased joint kinetics when fatigue occurs (Bini, Diefenthaeler, & Mota, 2010), and children adjust their power production during cycling when mass is added to their limbs (Brown & Jensen, 2006). The fact that children with CP were able to produce joint kinetics similar to TD peers after BTX treatment indicates that these children were able to perform the task more optimally due to a reduction in spasticity. The different task construction could be an indication that muscle control is more balanced and could be the reason the children were able to cycle longer at a certain target cadence than before BTX.

Besides the effects of BTX treatment on cycling itself, we were also interested in the impact of this treatment on the ability to adjust to changes in task demands. We used cadence as a modifier to investigate the influence of changes in movement speed on task adaptation. Joint kinetics were not influenced by changes in cadence pre and post BTX.

However, pre BTX, CP 1 had a substantial increase in jerk cost with an increase in cadence, while post BTX, no influence of cadence on jerk cost could be observed. Pre BTX, CP 2 had a similar cycling pattern as the TD peers. Nevertheless, there was still a reduction in jerk cost at 70 RPM post BTX. The substantial reduction in jerk cost at the higher cadences shows that BTX facilitated greater adaptation to changes in movement speed. The influence of cadence on jerk cost mediated by BTX is not surprising since spasticity affects the biomechanics of a locomotive skill more as speed increases (Van Campenhout, Bar-On, & Aertbeliën, 2014). Van Campenhout et al. found that spasticity in the GAS led to larger changes in kinematics and kinetics. For example, ankle angular velocity and ankle torques during loading response changed when children with CP increased their walking speed. In the same study, it was observed that spasticity in the HS changed the timing of the knee extension torque and the maximum value of the hip extension torque. Since spasticity in our study was reduced, the motion changed less at high speed than it did at pre BTX. As speed increased (not exceeding 80 RPM), the jerk cost was not influenced by an increase in cadence post BTX and was similar to what we observed during cycling of TD children (Study 1). Unfortunately, this does not explain why there was no cadence effect on the kinetics in our study.

In this study, jerk cost and joint kinematics and kinetics were shown to be useful measures to evaluate the effect of BTX on cycling. Even though the cycling technique was improved, no alterations were found in the asymmetry of bilateral work production. After the BTX injections, the children did not cycle until the post BTX cycling session. A decrease in spasticity can be observed within 12-72 hours after BTX injection, but the effects of this reduction in spasticity on the biomechanics of an individual may only be observed weeks later (Koman, Mooney, & Smith, 1993). The reduction in spasticity leads to improved ROM and increased muscle length, which gives the child with CP an opportunity to improve their locomotive skills using physical therapy. It is shown that gait parameters improve substantially more when the BTX injections are followed by highly intensive physical therapy (Scholtes, Dallmeijer, Knol, & Speth, 2007). The absence of cycling training after BTX injections in the current study could be the reason that the children with CP did not show an improvement in bilateral asymmetry. Further research is needed if intensive cycling therapy after BTX treatment is to lead to more improvement during cycling.

The children in this study underwent BTX as a part of their regular treatment, so we were unable to investigate the effect of BTX-mediated spasticity reduction of a particular muscle on a specific region of the crank cycle during cycling. Questions that would be interesting to study are: what is the effect of spasticity reduction in the GAS on the extension phase during cycling? Does it lead to a reduction in joint torques during the downstroke and to increased jerk cost of the crank cycle during the downstroke? Future research should investigate the relationship between specific muscles and the biomechanics in a specific phase of the cycling motion so that clinicians can better understand the effects of specific muscle treatment.

In conclusion, it was shown that BTX treatment is a potent intervention for improving cycling kinematics and kinetics in children with CP. Also, cycling biomechanics after BTX injections can be improved to approximately the same range as

TD peers. Post BTX, CP 1 and CP 2 cycled more fluently, with more typical phase-planes, lower peak joint torques, and had different timings for joint power production. The improved kinematics and joint kinetics indicated that the muscle control is more balanced, which gives the opportunity to strengthen the major muscle groups that are used for locomotive skills. The improved cycling fluency indicated that children in this were able to adapt better to changes in task demands making it easier for these children to increase their velocity during cycling training. Therefore, BTX can be useful to complement cycling therapy to provide the maximum therapeutic benefit to those children impacted by CP.

Chapter 5: General discussion

Motor development in children with cerebral palsy (CP) is often delayed in comparison to typically developing children (TD), due to a brain insult around birth (Paneth, 2008). TD children gradually learn how to interact with their environment, and many changes occur in their central nervous system and neuromuscular system.

Immediately after birth, TD children are incapable of producing voluntary, purposeful action. Within their first year of life, TD children learn to lift their head, sit, stand up and eventually walk. The ability to walk on two feet is the result of acquiring enough experience, strength, and coordination to be able to "stack the blocks" and overcome the gravity that is pulling on each block. For example, to be able to sit, infants need enough head, neck and trunk control to keep their upper body upright. From sitting, infants need to use trunk control in combination with the control of their lower extremities to stand up. During this challenging transition, the base of support shrinks from a wide round area to a small area that is defined by children's feet. At the same time as infants are learning how to balance their body in an upright position, the neuromuscular systems develop the ability to move limbs not with simply reflexive action or random flings but with reciprocal, coordinated movements. A combination of coordinated movement and the ability to overcome gravitational forces leads to the ability to perform complex locomotor skills, such as walking, running and cycling.

Unfortunately, not all children have the ability to combine locomotor skills and balance. Children with CP often produce disorganized muscle activation patterns during postural stability at a later age than TD children, which affects their balance (Woollacott et al., 1998). The differences in balance control of children with CP in comparison to TD children are due to changes in the central nervous system and changes in biomechanical posture. For example, standing in a crouching posture requires muscle activity to be adjusted to overcome gravity in contrast to what is required for standing up straight, because more mass is outside the base of support. When postural stability is affected in children with CP, locomotor skills, such as walking, are compromised (Woollacott & Shumway-Cook, 2005).

Impaired ambulation due to reduced walking skills leads to less physical activity in many children with CP, especially as their score on the Gross Motor Function Classification Scale (GMFCS) increases (Bjornson, Belza, Kartin, Logsdon, & McLaughlin, 2007). High levels of activity are necessary to reduce the chances of secondary changes that are related to CP, such as bone deformation and an affected cardiovascular system. Alternative forms of physical activity are often needed if an individual with CP has problems with coordination and balance that preclude walking and running (Damiano, 2006).

Cycling on a tricycle is one locomotor skill that is less dependent on postural control because only the upper body needs to be held upright. Furthermore, if trunk and head supports are mounted on the tricycle, the need for balance is minimal, and children with CP are often able to cycle, even if they are unable to sit upright (Fowler et al., 2007). This gives children with CP the opportunity to improve levels of physical activity, locomotor endurance, and muscle strength through cycling training (Fowler et al., 2010).

However, no standard protocol for cycling therapy in children with CP exists so far. For optimizing cycling training for the population with CP, we have to understand the biomechanics fully during cycling.

DISSERTATION OUTCOMES

The goal of this dissertation was to differentiate between age-related biomechanical changes in cycling and changes due to atypical development and to investigate the effect of spasticity reduction on such biomechanical changes. In Study 1, age-related differences in kinematics during upright cycling were studied to understand the development of cycling biomechanics in very young TD children. The outcome of this study revealed that 4-year-old TD children cycled with significantly more out-of-plane motion in comparison to older TD children and that jerk cost increased in cases of cycling at 100 revolutions per minute (RPM) or higher. These results indicate that cycling kinematics are stabilized by the age of 6-years.

In Study 2, the kinematics and kinetics of TD children and children with CP were observed during cycling on an ergometer with trunk support at different cadences. We investigated the abilities and limitations of children with CP during cycling to understand the most appropriate training regime during therapeutic cycling. Children with spastic CP cycled with more plantar flexion in the ankle, more out-of-plane motion, increased jerk cost and different pedal and joint kinetics. When cadence was increased, the children with CP had difficulties cycling fluently in comparison to TD children. Furthermore, the children less able to make task-specific force adaption, hence, they were limited in the range of cadences they were able to perform. We observed a distinction between different types of CP. Children with di/quadriplegia cycled successfully over a larger range of cadences but with more variability between the different cadence trials than children with hemiplegia. Furthermore, children with hemiplegic CP used their less affected leg as a force generator, which led to more asymmetry between their legs and more joint torques produced by their less affected leg. Because TD children have usually stabilized their cycling performance by the age of 6 years, we concluded that differences in kinematics and kinetics during cycling by children with CP were due to atypical development. As a result of this study, the question was raised, how would children with CP cycle differently if spasticity were reduced by botulinum-toxin (BTX) treatment?

In Study 3, we investigated the effects of spasticity reduction treatment by BTX injections on the cycling performances of 2 children with hemiplegic CP. In comparison to their pre BTX performances, these children cycled with fewer joint torques and reduced jerk cost 3 weeks after their treatment. Moreover, jerk cost was reduced to values similar to those produced by TD peers. The reduction in jerk cost indicated that the children with CP included in the study were more adaptable to changes in task demands. Similar to what we observed in Study 1 with TD children, jerk cost after BTX did not increase with cadence. No effect of BTX treatment was found on bilateral asymmetry.

CLINICAL APPLICATIONS

The results of these studies led us to conclude that the differences in the cycling biomechanics of young children with CP have important implications for cycling in a therapeutic setting.

1. Primary changes due to CP, such as spasticity and muscle weaknesses, were the reason that these children were less adaptable to changes in cadences and resistance, with the result that these children were only able to cycle at a small range of cadences. Upon reaching their upper limits of cadence, the physical demand was so high that cycling for longer periods of time at higher cadences was challenging. Therapists need to take into account that young children with CP should not undergo large changes in task demands when an intensive workload requiring high physical demand is not desirable.

2. The increased plantar flexion observed in these children led to adjustments in force applications to the pedals. Reducing plantar flexion during cycling would potentially lead to improvements in force application required to cycle more efficiently. The use of a dynamic ankle foot orthosis would help children with CP reduce plantar flexion and allow the ankle to move in a more nearly normal angle range.

3. Elevated joint torques in the frontal and transverse planes indicated that children with CP had more joint stress during cycling than their TD peers. However, during cycling at low cadences and low resistance, joint torques were lower than those reported during gait (Davids, Bagley, & Bryan, 1998; Novacheck & Gage, 2007). Therefore, carefully controlled therapeutic cycling is beneficial for young children with CP for increasing lower-extremity training with reduced occurrences of stress injuries.

4. The type of CP was a major factor in determining the ability of a child to perform a cycling task. Children with di/quadriplegic CP were more adaptable to changes in cadences than were children with hemiplegic CP; the former were able to cycle within a wider range of cadences. These findings present the opportunity to adjust cadences to alter the workload during cycling training. Children with di/quadriplegic CP cycled with more variability in biomechanics with increased joint torques, which shows that their muscle effort was elevated while cycling.

5. In contrast to children with di/quadriplegic CP, children with hemiplegic CP cycled successfully within a smaller range of cadences and with more bilateral asymmetry. The dominance of their less affected side during cycling suggests that muscles of their more affected side may not be efficiently trained during therapeutic cycling. Bilateral asymmetry was not influenced by BTX treatment, so post treatment the muscles in the more affected leg will still not be trained intensively.

6. Targeted BTX injections improved cycling fluency, reduced physical demand at higher cadences, and reduced joint stress in the legs of children with hemiplegic CP. These children's increased adaptability to changes in task demand indicates that, post treatment, children with hemiplegic CP can cycle longer than they could before treatment. The reduction in joint kinetics indicates that BTX treatment has great potential to reduce the risks of joint stress during cycling. In addition to benefitting children with hemiplegic CP, it is expected that BTX injections during cycling will also benefit children with spastic di-/quadriplegic CP, because cycling fluency and joint stress were affected by spasticity. The observed improvements in kinematics and kinetics during

cycling led us to conclude that BTX is an important supplement to cycling training in children with CP.

EXPECTED BENEFITS OF CYCLING THERAPY IN YOUNG CHILDREN WITH CP

Spasticity is one of the major symptoms of CP, and it greatly influences the performance of motor skills. The results of Study 3 indicate that cycling is affected by the influences of spasticity, because cycling biomechanics are more typical when spasticity is reduced by BTX treatment.

Exercise is commonly used as a treatment to reduce spasticity in children with CP, but there is no substantial evidence to support the effectiveness of this clinical strategy (Goldstein, 2001). We do not believe that cycling is beneficial for reducing spasticity. Nonetheless, encouraging cycling by young children is probably helpful in reducing other primary effects of CP, such as movement problems and reduced muscle strength. Due to the multi-segmental motion of cycling, which is similar to other locomotor skills, cycling is likely to enhance learning of several motor skills (Lauer, Johnston, Smith, & Lee, 2008).

Especially when children with CP train after BTX treatments, when their cycling patterns become more "typical," coordination between the major muscle groups will be more optimal. From earlier research, we know that older children (average 8.7 years) and adolescents with di/quadriplegic CP increase their muscle strength, endurance and functional skills through cycling training (Chen et al., 2012; Fowler et al., 2010; Williams

& Pountney, 2007). The cycling training in these studies increased duration, cadence, and resistance to enhance cycling performance in children with CP.

From the results of this dissertation research, we know that *young* children with hemi-, di- and quadriplegic CP are less able to adjust their cadences and resistance. A reduced capability to increase cadence and resistance leads to the expectation that improvement of muscle strength and endurance will take longer in young children with these conditions than in the older children and adolescents in previous studies. However, young children can cycle for longer periods of time at low cadences, which gives them opportunity to increase movement repetition for muscle coordination, which benefits their locomotor skills.

APPLICATION TO OTHER DISEASES

Considering that differences in the cycling biomechanics of children with CP are partly due to spasticity, it would seem likely that the effect of spasticity could be generalized to other disabilities, as spasticity is not a specific symptom of CP. Spasticity is found in children who suffer from disabilities, such as spinal cord injuries (Zidek & Srinivasan, 2003), spina bifida (Mazur, Stillwell, & Menelaus, 1986), stroke and traumatic brain injuries (Costeff, Groswasser, Landman, & Brenner, 1985). As with children with CP, if disabled children are capable of physical exercise and can move their legs, therapists have the option to increase children's physical activity with cycling. Therapeutic cycling in children with such disabilities is not as well studied. However, the practical implications for therapists discussed in this dissertation should be applicable for all disabilities with spasticity.

FUTURE RESEARCH

Two major questions have arisen from the research for this dissertation.

1. How are the individual muscles involved during cycling by children with CP capable of producing enough power to push the crank around, and what is the impact of increased cadence? The answer to this question will provide information about which muscles will be trained during cycling by young children with CP. The decrease in cycle fluency and the dragging of feet before bottom dead center indicate that children with CP are decelerating and accelerating the crank more to produce the same cadence output, which is the result of differences in motor control. Furthermore, the increased joint torques also indicate that children with CP rely on different muscles to produce power and that children with hemiplegic CP differ from children with di-/quadriplegia.

Muscle contribution can be predicted from net joint powers by static optimization. However, the problem with optimization is that the method cannot predict co-contraction among muscle around the same joint, and no estimation can be made about the contribution of the muscles to the acceleration of the segments and joints (Zajac, Neptune, & Kautz, 2002). This means that predictions based on static optimization would not be accurate in this population as co-contraction is increased in children with CP during cycling (Johnston, Barr, & Lee, 2007; Kaplan, 1995). A dynamical musculoskeletal model is needed that includes muscle-intrinsic properties, such as maximum isometric force, optimal fiber length, and activation and deactivation timings, to completely understand muscle coordination during cycling by children with and without CP. Therefore, research should attempt to estimate these muscle-intrinsic properties in these young populations and investigate how the muscles are activated to perform the cycling task.

2. Because children with CP engage in cycling training, it should be asked how their biomechanics are affected by such training. Cycling training is known to improve functional skills. One might speculate that cycling training would also improve kinematic and kinetic parameters during cycling. To address this question, we have also studied the effect of intensive cycling training sessions on one 6-, one 8- and one 9-year-old TD child (unpublished preliminary data). After these sessions, no measured improvement in jerk cost was observed. From Study 1, we learned that by the age of 6 years kinematic parameters are stabilized, so in hindsight, our negative findings are not surprising. However, children with CP have different kinematic and kinetic parameters, which may result in improvements after training. As proposed in Study 2, this training should start at low cadences and low resistance.

LIMITATIONS

The studies described in this dissertation had two principal limitations.

The children were asked to cycle at a pre-set target cadence. This design was developed to allow comparison between all children by assuring they were producing the same outcomes during a defined cycling task. However, there is the possibility that this design leads to more unnatural cycling performances. Children were required to stay within a specific range of cadence, and this might affect the fluency of their cycling performance. An inclusion of a self-determined cadence might inform us about a more natural cycling motion.

Another limitation was the pediatric ergometer that was used for this study. With this design, we were unable to alter the width between the pedals. Children with CP that produced "scissoring" (large hip adduction) were excluded from our study, due to their inability to sit on the ergometer and place their feet on the pedals. This limitation possibly led to an exclusion of a particular style of cycling performances undertaken by a subpopulation of children with CP.

SUMMARY

We investigated lower extremity kinematics and kinetics during stationary cycling. Three studies were performed to investigate age- and disability-related changes in cycling biomechanics. First, we showed that TD children were proficient cyclists by the age of 6-years. Second, the cycling performances of children with hemi-, di- and quadriplegic CP were characterized, and suggestions to increase the effectiveness of cycling therapy were made. Finally, the effect of BTX treatments on cycling performance was studied, and it was shown that BTX is an important treatment used in combination with cycling therapy. The results of these studies have practical implications for therapists and clinicians and lay a foundation for many exciting avenues of future research.

Appendices

APPENDIX A: INFORMED CONSENT

Consent for Participation in Research

Title: "Motor Control Differences between Typical Developed Children and Children with Cerebral Palsy or Autism"

Principal Investigator:	**Jody L. Jensen, Ph.D., Department of Kinesiology & Health Education, University of Texas at Austin, 512-232-2685**
Co-Investigators:	**Renate van Zandwijk Department of Kinesiology & Health Education, University of Texas at Austin, 512-232-2686**

Introduction

The purpose of this form is to provide you information that may affect your decision as to whether or not to allow your child to participate in this research study. The person performing the research will answer any of your questions. Read the information below and ask any questions you might have before deciding whether or not to take part. If you decide to be involved in this study, this form will be used to record your consent.

Purpose of the Study

Your child has been asked to participate in a research study about lower extremity muscle activity, specifically, the control of leg movements during stationary cycling. The purpose of this study is to investigate how children with a neurological disease control their legs different than typical developed children.

What will your child be asked to do?

If you agree to let your child participate in this study, he/she will be asked to visit the laboratory on one or two occasions. Your child will participate in three different kinds of tasks.

1. We will obtain your child's height, weight, and muscle assessment measurements.
2. We will ask your child to participate in a general assessment of movement skills (such as running, jumping, hopping);
3. We will ask your child to perform stationary cycling at different pedaling speeds and resistances. These speeds and resistances are similar to those experienced during normal cycling outdoors. We will take measurements of your child's leg to estimate your child's maximum workload. The resistances that your child will experience in this experiment will be no more than 20% of the maximum resistance that we estimate your child can cycle. The stationary bicycle is

adjustable such that for your child's height, the bicycle can be made comfortable for him or her to pedal. Simply, we can adjust the seat and handle bar position to suit your child. Testing should take no longer than two hours, including the time it takes to familiarize your child with the laboratory and to prepare for the cycling activity. If your child will be coming to the laboratory for multiple visits, plan on about an hour at the lab for each visit after the first day. Total pedaling time on any single day will be approximately 30 minutes.

To prepare for the collection of data, we may place reflective markers on your child's legs. These markers will go on his or her foot, ankle, and the outside of the knee and hip joints. Double-sided tape will keep these in place. We will record the activity of your child's muscles. In this case, sensors will be positioned on your child's legs. The sensors are attached with tape. The type of sensors we use is very common in this type of research and no problems have been reported following their use. The markers and sensors will not limit your child's movement. Your child will be able to pedal freely and comfortably. Pedaling will be constant for periods of 1 to 5 min, but well below your child's maximal effort. In one trial, we will ask your child to pedal at a self-selected maximum speed. This trial will be no more than 1 min in duration.

While your child pedals, special cameras will record the action of his or her lower limb. Only the reflective markers will be seen by these special cameras and he or she can in no way be identified from the camera images. Neither your child's name nor any personal information will be stored with the camera records. We may also record your child with a video camera while he or she is riding the bike.

This study will take approximately 2 hours of your time for one or two sessions and will include approximately 90 study participants.

Your child's participation will be video recorded.

What will you be asked to do?

1. We will ask you about your child's health history
2. We will ask you about your child's cycling experience and other sporting and recreational activities

What are the risks involved in this study?

This intervention may involve risks that are currently unforeseeable. Possible risks associated with this study are fatigue and muscle soreness. We are giving your child rest between every few minutes of cycling, but for some children this may not be sufficient. These symptoms typically dissipate within 48 hours. The stationary bicycle is very stable, and although the risk of falling is very small, the occurrence of such an incident is still possible. To mitigate the chance of falling, a safety frame has been constructed around the cycle to aid in support. In the unlikely event of injury as a result of your child's participation in this study, basic first aid will be provided at the time of injury and you will be encouraged to consult your physician. No treatment beyond initial first aid will be provided, and no payment can be provided in the event of a medical problem.

If you wish to discuss the information above or any other risks your child may experience, you may ask questions now or call the Principal Investigator listed on the front page of this form.

What are the possible benefits of this study?

Your child will receive no direct benefit from participating in this study; however, your child's participation may help us to better understand how children activate muscles and how we can be more effective in helping children acquire better movement skills through teaching or rehabilitation**.**

Do you have to participate?

No, your child's participation is voluntary. You may decide not to participate at all or, if you start the study, you may withdraw at any time. Withdrawal or refusing to participate will not affect your relationship with The University of Texas at Austin (University) in anyway.

If you would like to let your child participate, please return the signed forms to Dr. J.L. Jensen (jljensen@austin.utexas.edu) or Renate van Zandwijk (renate@utexas.edu). You will receive a copy of this form.

What are the alternatives to participating in this research?

Participation in this study is voluntary. You are making a decision as to whether you will or will not allow your child to participate. Your signature indicates that you have read the information provided above and have decided to allow your child to participate. You may withdraw your child at any time after you have signed this form should you choose to discontinue your involvement in this study. Withdrawing from the study will not affect your or your child's association with the University of Texas at Austin.

Will there be any compensation?

Your child will not receive any type of payment participating in this study.

How will your privacy and confidentiality be protected if you participate in this research study?

Your child's privacy and the confidentiality of your data will be protected by the use of a unique identification code for your child. Any information that is obtained in connection with this study and that can be identified with your child will remain confidential and will be disclosed only with your permission. Data collected via computer are stored in files that are labeled with codes, not names. Video records of your child's participation are also labeled with a unique identification code. If the results of this research are published or presented at scientific meetings, your child's identity will not be disclosed. All data are maintained indefinitely in this de-identified form and may be used in future research projects. All records related to your child's participation are stored in the Developmental Motor Control Laboratory – a secure location accessible to research personnel only by key and security code. The data will be added to a database we are building to catalogue the development of lower extremity control. This means that the data will not be destroyed.

If it becomes necessary for the Institutional Review Board to review the study records, information that can be linked to your child will be protected to the extent permitted by law. Your research records will not be released without your consent unless required by law or a court order. The data resulting from your child's participation may be made available to other researchers in the future for research purposes not detailed within this consent form. In these cases, the data will contain no identifying information that could associate it with you, or with your participation in any study.

If you choose to let your child participate in this study, your child will be video recorded. Any video recordings will be stored securely and only the research team will have access to the recordings. Recordings will be kept indefinitely, and added to a database that we are building to catalogue the development of lower extremity control.

Whom to contact with questions about the study?

Prior, during or after your child's participation you can contact the researcher Renate van Zandwijk at 512-232-2686 or send an email to renate@utexas.edu for any questions or if you feel that you have been harmed.

This study has been reviewed and approved by The University Institutional Review Board and the study number is **[STUDY NUMBER].**

Whom to contact with questions concerning your rights as a research participant?

For questions about your rights or any dissatisfaction with any part of this study, you can contact, anonymously if you wish, the Institutional Review Board by phone at (512) 471-8871 or email at orsc@uts.cc.utexas.edu.

Participation

If you agree to let your child participate, please return the signed forms to Dr. J.L. Jensen (jljensen@austin.utexas.edu) or Renate van Zandwijk (renate@utexas.edu).

Signature

Parent/Guardian of Study Participant: You have been informed about this study's purpose, procedures, possible benefits and risks, and you have received a copy of this form. You have been given the opportunity to ask questions before you sign, and you have been told that you can ask other questions at any time. You voluntarily agree to participate in this study. By signing this form, you are not waiving any of your legal rights.

______ I agree to be video recorded.

______ I do not want to be video recorded.

____

Printed Name

As a representative of this study, I have explained the purpose, procedures, benefits, and the risks involved in this research study.

____

Print Name of Person obtaining consent

__________________________________ __________________

Signature of Person obtaining consent Date

Study Participant Assent: You have been told about the purpose of this study, and told about what activities you will do if you take part in the study. You have been given the opportunity to ask questions before you sign this letter, and you have been told that you can ask other questions at any time. You voluntarily agree to participate in this study. And, you can stop participating at any time. Just tell the person in charge of this study that you would like to stop.

__________________________________ __________________

Signature of Study Participant Date

Photograph and Video Consent

I hereby give permission to use images or videos of my child for educational training, professional presentations, and/or professional publications. I understand that no explicit identifying information will accompany the presentation of pictures or videos, though the use of my child's image may lead to recognition of my child as a study participant.

____

Parent/Guardian Signature   Date

Appendix B: Physical Activity Questionnaire

ACTIVITY QUESTIONNAIRE – PARENT Subject ID:________

Please check any of the following activities that your child has done **AT LEAST 10 TIMES IN THE PAST 5 years.**

___ Aerobics ___ Band/Drill Team ___ Baseball

___ Basketball ___ Bowling ___ Cheerleading

___ Bike Riding ___ Dance classes ___ Football

___ Gymnastics ___ Hiking ___ Ice Skating

___ Roller Skating ___ Judo, Karate ___ Sailing

___ Running for exercise ___ Skateboarding ___ Snow Skiing

___ Soccer ___ Softball ___ Street Hockey

___ Swimming training ___ Tennis ___ Volleyball

___ Water Skiing ___ Weight Training ___ Wrestling

For each of the activities above, please fill in the activity log where possible

Activity	Number of Years	Months / Year	Weeks / Month	Days / Weekday	Days / Weekend	Hours / Weekday	Hours / Weekend	TOTAL HOURS
e.g. Sailing	2	6	4 (all)	3	1	1	2	240

How does your child get **to** school?
a. Car b. Catches the bus
c. Walk c. Rides a bike

How does your child get home **from** school?
a. Car b. Catches the bus
c. Walk c. Rides a bike

If walking or riding, how far is it to/from school? ___________________________

How many hours **per weekday** does your child watch Television and videos, play computer games, and surf the internet before and after school?

a. None b. 1 hour or less c. 2 to 3 hours
d. 3 to 5 hours e. 5 hours or more

How many hours **per weekend** does your child watch Television and videos, play computer games, and surf the internet?

a. None b. 1 hour or less c. 2 to 3 hours
d. 3 to 5 hours e. 5 hours or more

Has your child participated in any sport **races or competitions**?
____ yes ____ no

If so, please provide details:

____

____

____

____

At what age did your child start riding a bike?_________________

Does your child ride a bike to get around (e.g. Does your child ride his/her bike to school/friends?) Please explain.

____

____

Does your child own a bike? _______________

How often has your child ridden a bike in the past five years?

Activity	**Number of Years**	**Months / Year**	**Weeks / Month**	**Days / Weekday**	**Days / Weekend**	**Hours / Weekday**	**Hours / Weekend**	**TOTAL HOURS**
e.g.	2	6	4 (all)	3	1	1	2	240
Biking								

If your child does not ride a bike on a regular basis, please provide specific information on how often and how many hours your child has ridden a bike within the past 5 years.

____

____

____

____

If there are other things about your child's cycling history that you think are worth mentioning, please write them down here.

____

APPENDIX C: CHECKLIST STUDY 1

Subject: ____________________
Date: ________________________

Preparation

- Reserve parking pass and pick it up
- Get Informed Consent and activity forms
- Get order cycling speeds
- Calibrate cameras and G=get markers ready (16 markers +16 EMG)

During Session

- Sign Informed Consent + fill in questionnaire
- BOT
- Length ___________ and weight ______________
- Predict peak power
- Attach markers
- Take short trial while participant stands
- Calibrate pedals
- Measure body angles on bike
- Seat height: __________ and crank length _________
- Static Calibration

APPENDIX D: ESTIMATING PEAK POWER

	A	B	C	D
1	Subject ID			
2	Thigh Length 1 - proximal	D2/2	Thigh length	
3	Thigh Length 2 - distal	D2/2		
4	**Circumferences**		**Lean limb circumferences**	
5	C1 (gluteal furrow)		LC1	=B5-B15*PI()
6	C2 (mid thigh)		LC2	=B6-B22*PI()
7	C3 (proximal patella)		LC3	=B7-B29*PI()
8	**Skin fold thickness**		**Lean limb diameters**	
9	gluteal furrow front 1		LD1	D5/PI()
10	gluteal furrow front 2		LD2	D6/PI()
11	gluteal furrow front 3		LD3	D7/PI()
12	gluteal furrow back 1		Radius	
13	gluteal furrow back 2		R1	D9/2
14	gluteal furrow back 3		R2	D10/2
15	**average gluteal furrow**	=AVERAGE(B9:B14)	R3	D11/2
16	mid thigh front 1		**Heights**	
17	mid thigh front 2		height1(proximal)	=SQRT((B2)^2-(D14-D13)^2)
18	mid thigh front 3		height2 (distal)	=SQRT((B3)^2-(D15-D14)^2)
19	mid thigh back 1		**Volumes**	
20	mid thigh back 2		Cone1 (proximal)	=(PI()*D17/3)*(D13^2+D14*D13+D14^2)
21	mid thigh back 3		Cone2 (distal)	=(PI()*D18/3)*(D14^2+D15*D14+D15^2)
22	**average mid thigh**	=AVERAGE(B16:B21)	**Sum**	=(D20+D21)/1000
23	proximal patella front 1		**Pmax**	=(215.96*D22+48.53
24	proximal patella front 2			
25	proximal patella front 3			
26	proximal patella back 1			
27	proximal patella back 2			
28	proximal patella back 3			
29	**average proximal patella**	=AVERAGE(B23:B28)		

APPENDIX E: BOT-2 SHORT FORM

SHORT Form

Point Score

Subtest 1: Fine Motor Precision

Raw Score

3 Drawing Lines through Paths—Crooked — errors

Raw	≥21	15–20	10–14	6–9	4–5	2–3	1	0
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**

6 Folding Paper — points

Raw	0	1–2	3–4	5–6	7–8	9–10	11	12
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**

Subtest 2: Fine Motor Integration

	Basic Shape	Closure	Edges	Orientation	Overlap	Overall Size	Raw Score*
2 Copying a Square	0 1	0 1	0 1	0 1		0 1	points
7 Copying a Star	0 1	0 1	0 1	0 1		0 1	points

Subtest 3: Manual Dexterity

Raw Score: Trial 1, Trial 2

2 Transferring Pennies — pennies, pennies

Raw	0–2	3–4	5–6	7–8	9–10	11–12	13–14	15–16	17–18	19–20
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**

Subtest 4: Bilateral Coordination

Raw Score: Trial 1, Trial 2

3 Jumping in Place—Same Sides Synchronized — jumps, jumps

Raw	0	1	2–4	5
Point	**0**	**1**	**2**	**3**

6 Tapping Feet and Fingers—Same Sides Synchronized — taps, taps

Raw	0	1	2–4	5–9	10
Point	**0**	**1**	**2**	**3**	**4**

Subtest 5: Balance

Raw Score: Trial 1, Trial 2

2 Walking Forward on a Line — steps, steps

Raw	0	1–2	3–4	5	6
Point	**0**	**1**	**2**	**3**	**4**

7 Standing on One Leg on a Balance Beam—Eyes Open — seconds, seconds

Raw	0.0–0.9	1.0–2.9	3.0–5.9	6.0–9.9	10
Point	**0**	**1**	**2**	**3**	**4**

Subtest 6: Running Speed and Agility

Raw Score: Trial 1, Trial 2

3 One-Legged Stationary Hop — hops, hops

Raw	0	1–2	3–5	6–9	10–14	15–19	20–24	25–29	30–39	40–49	≥50
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**

Subtest 7: Upper-Limb Coordination

Raw Score: Trial 1, Trial 2

1 Dropping and Catching a Ball—Both Hands — catches

Raw	0	1	2	3	4	5
Point	**0**	**1**	**2**	**3**	**4**	**5**

6 Dribbling a Ball—Alternating Hands — dribbles, dribbles

Raw	0	1	2	3	4–5	6–7	8–9	10
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**

Subtest 8: Strength

Raw Score

2a Knee Push-ups *OR* (circle one) **2b** Full Push-ups — push-ups

Raw	0	1–2	3–5	6–10	11–15	16–20	21–25	26–30	31–35	≥36
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**

3 Sit-ups — sit-ups

Raw	0	1–2	3–5	6–10	11–15	16–20	21–25	26–30	31–35	≥36
Point	**0**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**

Notes & Observations

Total Point Score Short Form (max = 88)

* For Subtest 2: Fine Motor Integration, add the facet scores, record the sum in the Raw Score column, and transfer the raw score for each item directly to the corresponding oval in the Point Score column.

8

APPENDIX F: QUESTIONNAIRE CEREBRAL PALSY

Questionnaire CP background Subject: ___________________

What type of CP is your child diagnosed with (spastic, ataxic, dystonic)? ____________

What subtype of CP is your child diagnosed with monoplegia (one limb), hemiplegia (both arm and leg on same side), paraplegia or diplegia (both legs) or quadriplegia (both arms and legs involved)? ______________________________

Which side of the body has the most spastic? _____________________________

When was your child diagnosed? ______________________________________

Do the doctors have an idea what the cause was and when it occurred? ____

Did you child had any of the following treatments?

	Yes	No
Selective dorsal rhizotomy	◻	◻
If yes, when? ____		
Baclofen Pump	◻	◻
Oral medication	◻	◻
If yes, which? ____		
Orthopedic surgery	◻	◻
If yes, what? ____		

Botox injections ☐ ☐

If yes, when was the last injection? ________________________________

Is your child classified by the doctor on a specific scale, such as GMFCS or GMFM? _________

If yes, which scale and which level?_______________________________

Are there any other things we should know? ______________________________

____

____

Questions to determine GMFCS-level (2-3 years old)

Sitting

Sits on the floor without support ___________

Sits on the floor without support, but has problems when is manipulating objects: ___

Moves in and out of sitting on the floor without assistance of adults ___________

Sits on the floor often by "W-sitting" (sitting between flexed and internally rotated hips and knees) ___________

Requires adult assistance to assume sitting on the floor ___________

Sits on the floor when placed, but are unable to maintain alignment and balance without use of their hands for support ___________

Needs adaptive assistant for sitting ___________

Crawling

Self-mobility for short distances (within a room) is achieved through rolling, creeping on stomach, or crawling on hands and knees without reciprocal leg movement __________

Creeps on their stomach or crawls on hands and knees (often without reciprocal leg movements) as their primary methods of self-mobility ___________

Crawl on hands and knees with a reciprocal pattern ___________

Standing, Cruising and Walking

Can pull to stand on a stable surface ___________

Cruises holding onto furniture ___________

Needs adaptive assistant for standing ___________

Walks independent and choose this as preferred method of mobility ___________

Walks using an assistive mobility device as preferred method of mobility ___________

Children may walk short distances indoors using a hand-held mobility device (walker) and adult assistance for steering and turning. ___________

Questions to determine GMFCS-level (4-5 years old)

Sitting

Gets into and out of, and sit in, a chair without the need for hand support ___________

Sits in a chair with both hands free to manipulate objects ___________

Sits on a regular chair but may require pelvic or trunk support to maximize hand function ____

Sits on a chair but need adaptive seating for trunk control and to maximize hand function ___

Moves in and out of chair sitting using a stable surface to push on or pull up with their arms ___________

Standing and Cruising

Moves from the floor and from chair sitting to standing without the need for objects for support ___________

Moves from the floor to standing and from chair sitting to standing but often require a stable surface to push or pull up on with their arms ___________

Walking and other locomotor skills

Walks indoors and outdoors independently ___________

Walks with a hand-held mobility device on level surfaces and climb stairs with assistance from an adult ___________

Frequently are transported when traveling for long distances or outdoors on uneven terrain __

May at best walk short distances with a walker and adult supervision but has difficulty turning and maintaining balance on uneven surfaces ___________

Is transported in the community or may achieve self-mobility using a powered wheelchair ___________

Climbs stairs independently and emerging ability to run and jump ___________

Climbs stairs holding onto a railing but are unable to run or jump ___________

Questions to determine GMFCS-level (6-11 years old)

Sitting

Sits independently ___________

When seated, may requires a seat belt for pelvic alignment and balance ___________

Sit-to-stand and floor-to-stand transfers require physical assistance of a person or support surface ___________

Requires adaptive seating for trunk and pelvic control and physical assistance for most transfers ___________

Walking and transportation

Walks in most settings ___________

Walks at home, school, outdoors, and in the community ___________

Is able to walk up and down curbs without physical assistance ___________

May experiences difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas, confined spaces or when carrying objects ___________

Outdoors and in the community, may walks with physical assistance, a hand-held mobility device, or use wheeled mobility when traveling long distances ___________

Walks using a hand-held mobility device in most indoor settings ___________

When traveling long distances, uses some form of wheeled mobility ___________

Uses methods of mobility that require physical assistance or powered mobility in most settings ___________

At home, children use floor mobility (roll, creep, or crawl), walk short distances with physical assistance, or use powered mobility ___________

When positioned, children may use a body support walker at home or school _________

At school, outdoors, and in the community, children are transported in a manual wheelchair or use powered mobility ___________

Other locomotor skills

Is able to walk up and down the stairs without the use of a railing ___________

Walks up and down stairs holding onto a railing or with physical assistance if there is no railing ___________

Perform gross motor skills such as running and jumping but speed, balance, and coordination are limited ___________

Has at best only minimal ability to perform gross motor skills such as running and jumping ___________

May participates in physical activities and sports depending on personal choices and environmental factors ___________

Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports ___________

Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility ___________

APPENDIX G: CHECKLIST STUDY 2 AND 3

Subject: ____________________
Date: ________________________

Preparation

- Reserve parking pass and pick it up
- Get Informed Consent and questionnaires
- Get order cycling speeds
- Calibrate cameras and Get markers ready (16 markers +16 EMG)
-

During Session

- Sign Informed Consent + fill in questionnaire
- Length ___________ and weight ______________
- Leg Circumference

Leg Circumference	Distance between
1. ________	1. ________
2. ________	2. ________
3. ________	3. ________
4. ________	4. ________
5. ________	5. ________
6. ________	6. ________
7. ________	7. ________
8. ________	

- Modified Ashworth Scale
  1. Hip flexor left __ Hip flexor right __
  2. Hip adductor left ___ Hip adductor right ___
  3. RF left ___ RF right ___
  4. Hamstring left ___ Hamstring right ___
  5. Gastroc left ___ Gastroc right ___
  6. Soleus left ___ Soleus right ___
  7. Ankle plantar flexion left __ Ankle plantar flexion right ___

- Attach markers
- Take short trial while participant stands
- Calibrate pedals
- Measure body angles on bike
- Seat height: __________ and crank length _________
- Static Calibration

APPENDIX H: GROSS MOTOR FUNCTION CLASSIFICATION SYSTEM

CanChild Centre for Childhood Disability Research

INTRODUCTION & USER INSTRUCTIONS

The Gross Motor Function Classification System (GMFCS) for cerebral palsy is based on self-initiated movement, with emphasis on sitting, transfers, and mobility. When defining a five-level classification system, our primary criterion has been that the distinctions between levels must be meaningful in daily life. Distinctions are based on functional limitations, the need for hand-held mobility devices (such as walkers, crutches, or canes) or wheeled mobility, and to a much lesser extent, quality of movement. The distinctions between Levels I and II are not as pronounced as the distinctions between the other levels, particularly for infants less than 2 years of age.

The expanded GMFCS (2007) includes an age band for youth 12 to 18 years of age and emphasizes the concepts inherent in the World Health Organization's International Classification of Functioning, Disability and Health (ICF). We encourage users to be aware of the impact that **environmental** and **personal** factors may have on what children and youth are observed or reported to do. The focus of the GMFCS is on determining which level best represents the **child's or youth's present abilities and limitations in gross motor function**. Emphasis is on usual **performance** in home, school, and community settings (i.e., what they do), rather than what they are known to be able to do at their best (capability). It is therefore important to classify current performance in gross motor function and not to include judgments about the quality of movement or prognosis for improvement.

The title for each level is the method of mobility that is most characteristic of performance after 6 years of age. The descriptions of functional abilities and limitations for each age band are broad and are not intended to describe all aspects of the function of individual children/youth. For example, an infant with hemiplegia who is unable to crawl on his or her hands and knees, but otherwise fits the description of Level I (i.e., can pull to stand and walk), would be classified in Level I. The scale is ordinal, with no intent that the distances between levels be considered equal or that children and youth with cerebral palsy are equally distributed across the five levels. A summary of the distinctions between each pair of levels is provided to assist in determining the level that most closely resembles a child's/youth's current gross motor function.

We recognize that the manifestations of gross motor function are dependent on age, especially during infancy and early childhood. For each level, separate descriptions are provided in several age bands. Children below age 2 should be considered at their corrected age if they were premature. The descriptions for the 6 to 12 year and 12 to18 year age bands reflect the potential impact of environment factors (e.g., distances in school and community) and personal factors (e.g., energy demands and social preferences) on methods of mobility.

An effort has been made to emphasize abilities rather than limitations. Thus, as a general principle, the gross motor function of children and youth who are able to perform the functions described in any particular level will probably be classified at or above that level of function; in contrast, the gross motor function of children and youth who cannot perform the functions of a particular level should be classified below that level of function.

OPERATIONAL DEFINITIONS

Body support walker – A mobility device that supports the pelvis and trunk. The child/youth is physically positioned in the walker by another person.

Hand-held mobility device – Canes, crutches, and anterior and posterior walkers that do not support the trunk during walking.

Physical assistance – Another person manually assists the child/youth to move.

Powered mobility – The child/youth actively controls the joystick or electrical switch that enables independent mobility. The mobility base may be a wheelchair, scooter or other type of powered mobility device.

Self-propels manual wheelchair – The child/youth actively uses arms and hands or feet to propel the wheels and move.

Transported – A person manually pushes a mobility device (e.g., wheelchair, stroller, or pram) to move the child/youth from one place to another.

Walks – Unless otherwise specified indicates no physical assistance from another person or any use of a hand-held mobility device. An orthosis (i.e., brace or splint) may be worn.

Wheeled mobility – Refers to any type of device with wheels that enables movement (e.g., stroller, manual wheelchair, or powered wheelchair).

GENERAL HEADINGS FOR EACH LEVEL

LEVEL I **-** Walks without Limitations

LEVEL II **-** Walks with Limitations

LEVEL III **-** Walks Using a Hand-Held Mobility Device

LEVEL IV **-** Self-Mobility with Limitations; May Use Powered Mobility

LEVEL V **-** Transported in a Manual Wheelchair

DISTINCTIONS BETWEEN LEVELS

Distinctions Between Levels I and II - Compared with children and youth in Level I, children and youth in Level II have limitations walking long distances and balancing; may need a hand-held mobility device when first learning to walk; may use wheeled mobility when traveling long distances outdoors and in the community; require the use of a railing to walk up and down stairs; and are not as capable of running and jumping.

Distinctions Between Levels II and III - Children and youth in Level II are capable of walking without a hand-held mobility device after age 4 (although they may choose to use one at times). Children and youth in Level III need a hand-held mobility device to walk indoors and use wheeled mobility outdoors and in the community.

Distinctions Between Levels III and IV - Children and youth in Level III sit on their own or require at most limited external support to sit, are more independent in standing transfers, and walk with a hand-held mobility device. Children and youth in Level IV function in sitting (usually supported) but self-mobility is limited. Children and youth in Level IV are more likely to be transported in a manual wheelchair or use powered mobility.

Distinctions Between Levels IV and V - Children and youth in Level V have severe limitations in head and trunk control and require extensive assisted technology and physical assistance. Self-mobility is achieved only if the child/youth can learn how to operate a powered wheelchair.

BEFORE 2nd BIRTHDAY

LEVEL I: Infants move in and out of sitting and floor sit with both hands free to manipulate objects. Infants crawl on hands and knees, pull to stand and take steps holding on to furniture. Infants walk between 18 months and 2 years of age without the need for any assistive mobility device.

LEVEL II: Infants maintain floor sitting but may need to use their hands for support to maintain balance. Infants creep on their stomach or crawl on hands and knees. Infants may pull to stand and take steps holding on to furniture.

LEVEL III: Infants maintain floor sitting when the low back is supported. Infants roll and creep forward on their stomachs.

LEVEL IV: Infants have head control but trunk support is required for floor sitting. Infants can roll to supine and may roll to prone.

LEVEL V: Physical impairments limit voluntary control of movement. Infants are unable to maintain antigravity head and trunk postures in prone and sitting. Infants require adult assistance to roll.

BETWEEN 4th AND 6th BIRTHDAY

LEVEL I: Children get into and out of, and sit in, a chair without the need for hand support. Children move from the floor and from chair sitting to standing without the need for objects for support. Children walk indoors and outdoors, and climb stairs. Emerging ability to run and jump.

LEVEL II: Children sit in a chair with both hands free to manipulate objects. Children move from the floor to standing and from chair sitting to standing but often require a stable surface to push or pull up on with their arms. Children walk without the need for a hand- held mobility device indoors and for short distances on level surfaces outdoors. Children climb stairs holding onto a railing but are unable to run or jump.

LEVEL III: Children sit on a regular chair but may require pelvic or trunk support to maximize hand function. Children move in and out of chair sitting using a stable surface to push on or pull up with their arms. Children walk with a hand-held mobility device on level surfaces and climb stairs with assistance from an adult. Children frequently are transported when traveling for long distances or outdoors on uneven terrain.

LEVEL IV: Children sit on a chair but need adaptive seating for trunk control and to maximize hand function. Children move in and out of chair sitting with assistance from an adult or a stable surface to push or pull up on with their arms. Children may at best walk short distances with a walker and adult supervision but have difficulty turning and maintaining balance on uneven surfaces. Children are transported in the community. Children may achieve self-mobility using a powered wheelchair.

LEVEL V: Physical impairments restrict voluntary control of movement and the ability to maintain antigravity head and trunk postures. All areas of motor function are limited. Functional limitations in sitting and standing are not fully compensated for through the use of adaptive equipment and assistive technology. At Level V, children have no means of independent movement and are transported. Some children achieve self-mobility using a powered wheelchair with extensive adaptations.

BETWEEN 6th AND 12th BIRTHDAY

LEVEL I: Children walk at home, school, outdoors, and in the community. Children are able to walk up and down curbs without physical assistance and stairs without the use of a railing. Children perform gross motor skills such as running and jumping but speed, balance, and coordination are limited. Children may participate in physical activities and sports depending on personal choices and environmental factors.

LEVEL II: Children walk in most settings. Children may experience difficulty walking long distances and balancing on uneven terrain, inclines, in crowded areas, confined spaces or when carrying objects. Children walk up and down stairs holding onto a railing or with physical assistance if there is no railing. Outdoors and in the community, children may walk with physical assistance, a hand-held mobility device, or use wheeled mobility when traveling long distances. Children have at best only minimal ability to perform gross motor skills such as running and jumping. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports.

LEVEL III: Children walk using a hand-held mobility device in most indoor settings. When seated, children may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance of a person or support surface. When traveling long distances, children use some form of wheeled mobility. Children may walk up and down stairs holding onto a railing with supervision or physical assistance. Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility.

LEVEL IV: Children use methods of mobility that require physical assistance or powered mobility in most settings. Children require adaptive seating for trunk and pelvic control and physical assistance for most transfers. At home, children use floor mobility (roll, creep, or crawl), walk short distances with physical assistance, or use powered mobility. When positioned, children may use a body support walker at home or school. At school, outdoors, and in the community, children are transported in a manual wheelchair or use powered mobility. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.

LEVEL V: Children are transported in a manual wheelchair in all settings. Children are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and and/or mobility but limitations are not fully compensated by equipment. Transfers require complete physical assistance of an adult. At home, children may move short distances on the floor or may be carried by an adult. Children may achieve self- mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

BETWEEN 12th AND 18th BIRTHDAY

LEVEL I: Youth walk at home, school, outdoors, and in the community. Youth are able to walk up and down curbs without physical assistance and stairs without the use of a railing. Youth perform gross motor skills such as running and jumping but speed, balance, and coordination are limited. Youth may participate in physical activities and sports depending on personal choices and environmental factors.

LEVEL II: Youth walk in most settings. Environmental factors (such as uneven terrain, inclines, long distances, time demands, weather, and peer acceptability) and personal preference influence mobility choices. At school or work, youth may walk using a hand- held mobility device for safety. Outdoors and in the community, youth may use wheeled mobility when traveling long distances. Youth walk up and down stairs holding a railing or with physical assistance if there is no railing. Limitations in performance of gross motor skills may necessitate adaptations to enable participation in physical activities and sports.

LEVEL III: Youth are capable of walking using a hand-held mobility device. Compared to individuals in other levels, youth in Level III demonstrate more variability in methods of mobility depending on physical ability and environmental and personal factors. When seated, youth may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance from a person or support surface. At school, youth may self-propel a manual wheelchair or use powered mobility. Outdoors and in the community, youth are transported in a wheelchair or use powered mobility. Youth may walk up and down stairs holding onto a railing with supervision or physical assistance. Limitations in walking may necessitate adaptations to enable participation in physical activities and sports including self-propelling a manual wheelchair or powered mobility.

LEVEL IV: Youth use wheeled mobility in most settings. Youth require adaptive seating for pelvic and trunk control. Physical assistance from 1 or 2 persons is required for transfers. Youth may support weight with their legs to assist with standing transfers. Indoors, youth may walk short distances with physical assistance, use wheeled mobility, or, when positioned, use a body support walker. Youth are physically capable of operating a powered wheelchair. When a powered wheelchair is not feasible or available, youth are transported in a manual wheelchair. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports, including physical assistance and/or powered mobility.

LEVEL V: Youth are transported in a manual wheelchair in all settings. Youth are limited in their ability to maintain antigravity head and trunk postures and control arm and leg movements. Assistive technology is used to improve head alignment, seating, standing, and mobility but limitations are not fully compensated by equipment. Physical assistance from 1 or 2 persons or a mechanical lift is required for transfers. Youth may achieve self-mobility using powered mobility with extensive adaptations for seating and control access. Limitations in mobility necessitate adaptations to enable participation in physical activities and sports including physical assistance and using powered mobility.

APPENDIX I: MODIFIED ASHWORTH SCALE INSTRUCTIONS

General Information (derived Bohannon and Smith, 1987):

- Place the patient in a supine position
- If testing a muscle that primarily flexes a joint, place the joint in a maximally flexed position and move to a position of maximal extension over one second (count "one thousand one")
- If testing a muscle that primarily extends a joint, place the joint in a maximally extended position and move to a position of maximal flexion over one second (count "one thousand one")
- Score based on the classification below

Scoring (taken from Bohannon and Smith, 1987):

0	No increase in muscle tone
1	Slight increase in muscle tone, manifested by a catch and release or by minimal resistance at the end of the range of motion when the affected part(s) is moved in flexion or extension
1+	Slight increase in muscle tone, manifested by a catch, followed by minimal resistance throughout the remainder (less than half) of the ROM
2	More marked increase in muscle tone through most of the ROM, but affected part(s) easily moved
3	Considerable increase in muscle tone, passive movement difficult
4	Affected part(s) rigid in flexion or extension

Patient Instructions:

The patient should be instructed to relax.

Modified Ashworth Scale Testing Form

Name:____ Date:___________

Muscle	Tested Score
____________________	______
____________________	______
____________________	______
____________________	______
____________________	______
____________________	______
____________________	______
____________________	______
____________________	______

<u>Reference for test instructions:</u>

Bohannon, R. and Smith, M. (1987). "Interrater reliability of a modified Ashworth scale of muscle spasticity." Physical Therapy 67(2): 206.

Test instructions provided courtesy of Richard Bohannon PT, PhD and Melissa Smith, PT

APPENDIX J: JERK COST

Jerk cost is a measurement of fluency and movements are considered fluent if the accelerative transients are kept to a minimum (Nelson, 1983).

Jerk is the third derivative of position (Table 1). A fluent motion can be observed if the sum of the squared jerk along a trajectory (jerk cost) is minimized.

<u>Reason for large unit:</u>

When jerk is squared, the unit of jerk (deg/s^3) becomes deg^2/s^6. Taking the integral from this function will lead to the unit for jerk cost (deg/s^5).

Table A1: calculation time dependent variables

Variable	Calculation	Unit
Angle (θ)	dθ(t)	Deg
Angular velocity (ω)	dθ(t)/dt	Deg/s
Angular acceleration (α)	dω(t)/dt	Deg/s^2
Jerk (J)	dα(t)/dt	Deg/s^3
Jerk Cost	$1/2 \int_0^T J(t)^2 dt$	Deg^2/s^5

Reference

Nelson, W. L. (1983). Physical principles for economies of skilled movements. *Biological Cybernetics*, *46*(2), 135–147.

APPENDIX K: JOINT ANGLES IN STUDY 1

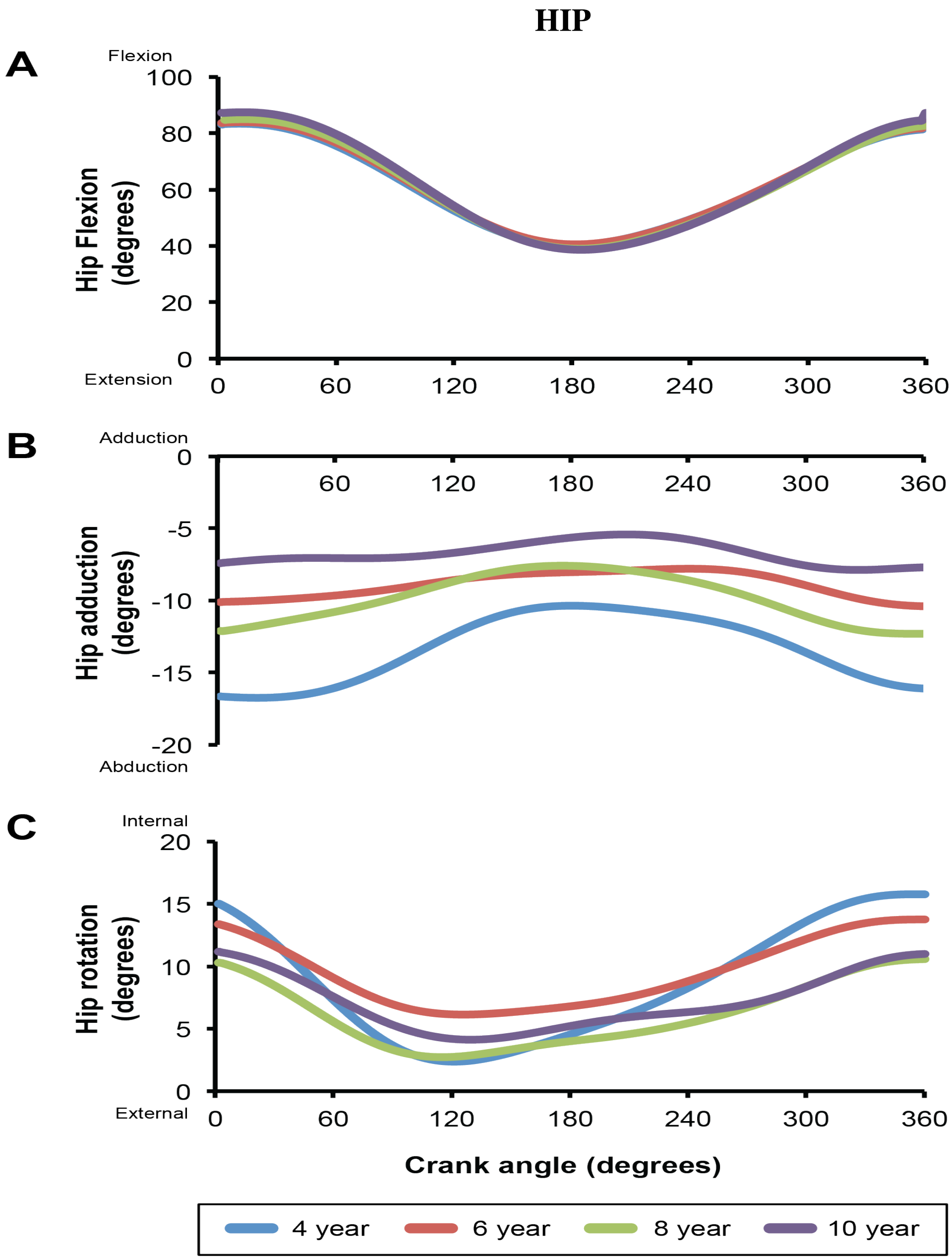


Figure A1: Hip motion: flexion (A), adduction (B) and rotation (C).

KNEE

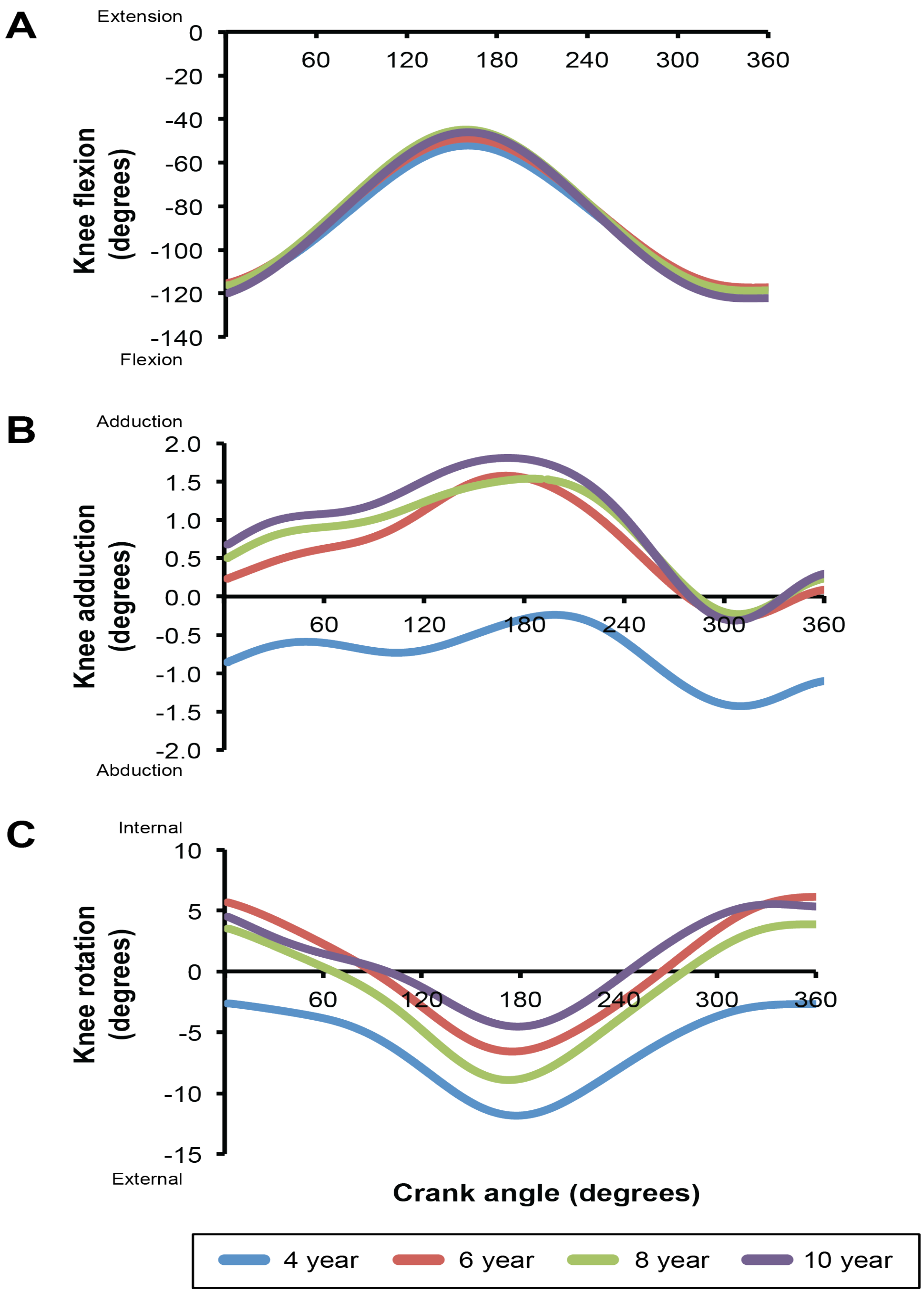


Figure A2: Knee motion: flexion (A), adduction (B) and rotation (C).

ANKLE

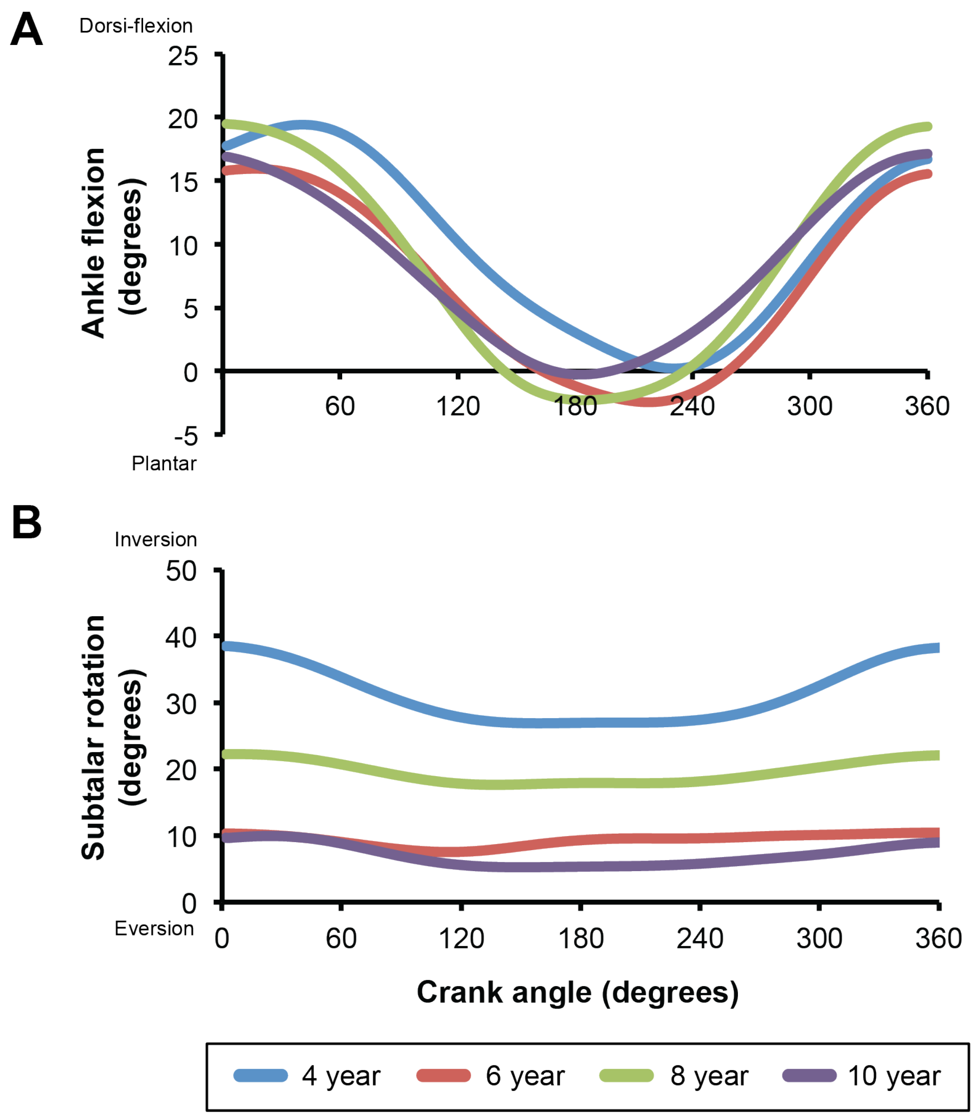


Figure A3: Ankle motion: ankle flexion (A) and subtalar rotation. (B)

APPENDIX L: JOINT ANGLES AT 30 RPM IN STUDY 2

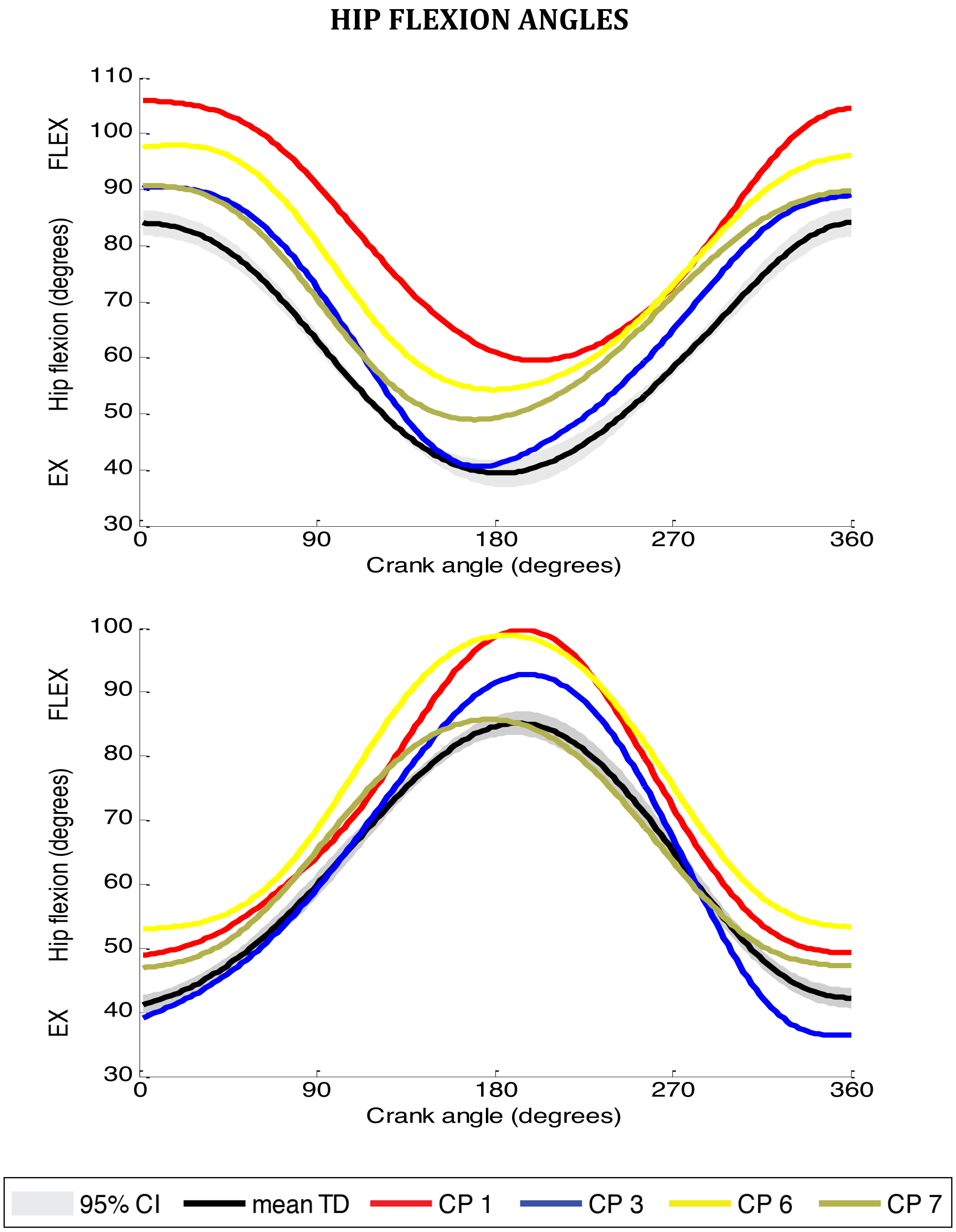


Figure A4: Hip flexion angles at 30 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION ANGLES

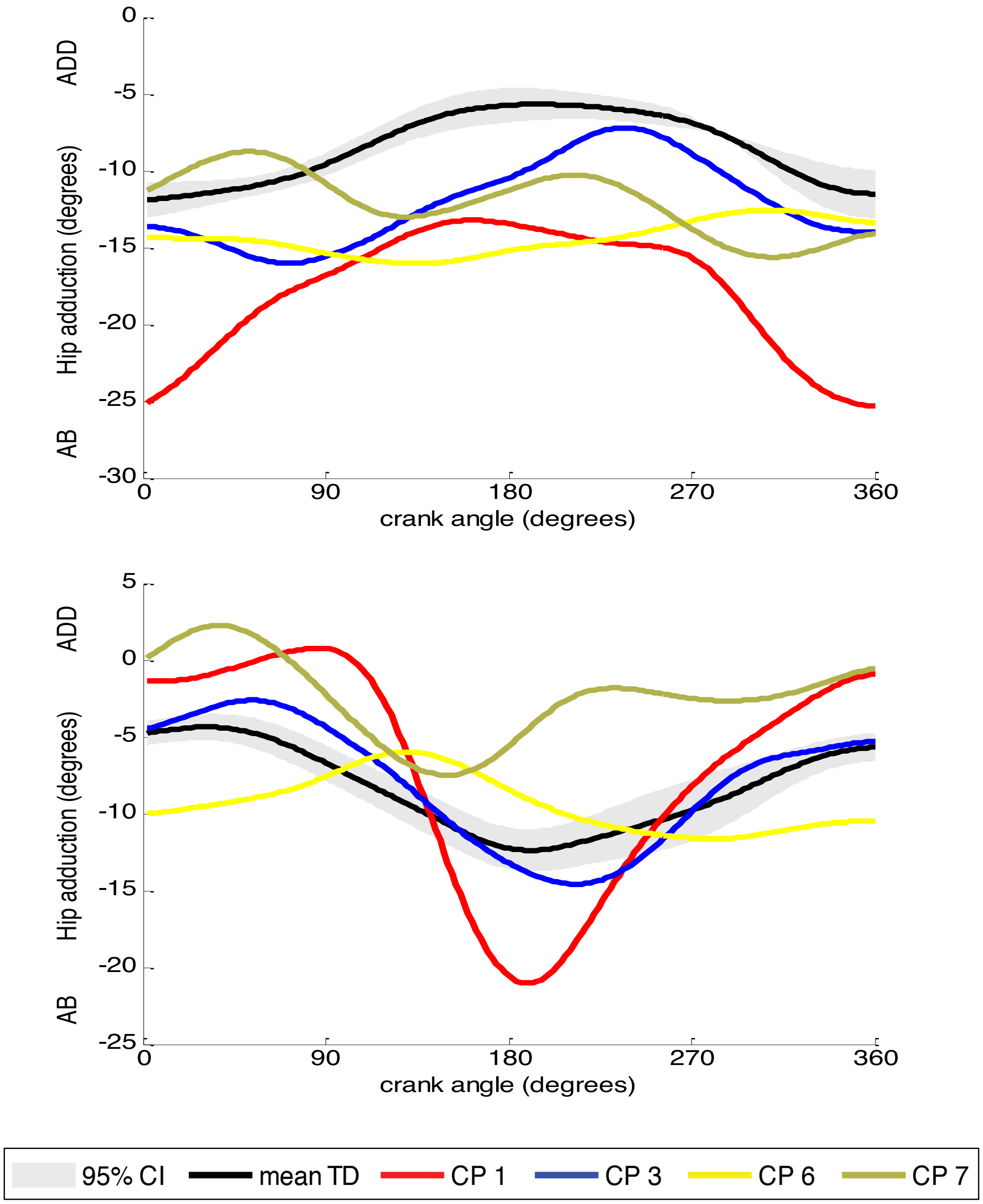


Figure A5: Hip adduction angles at 30 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION ANGLES

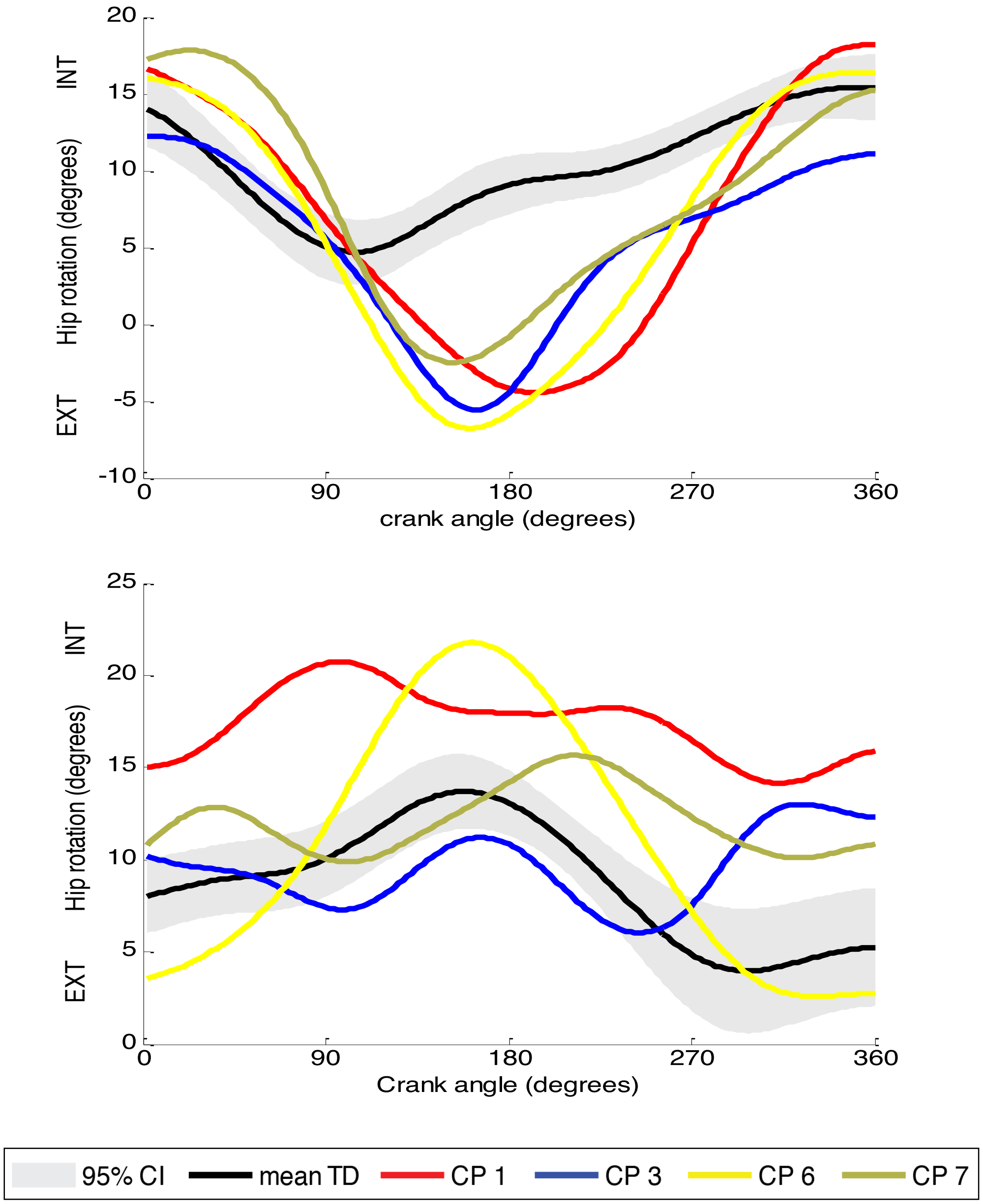


Figure A6: Hip rotation angles at 30 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION ANGLES

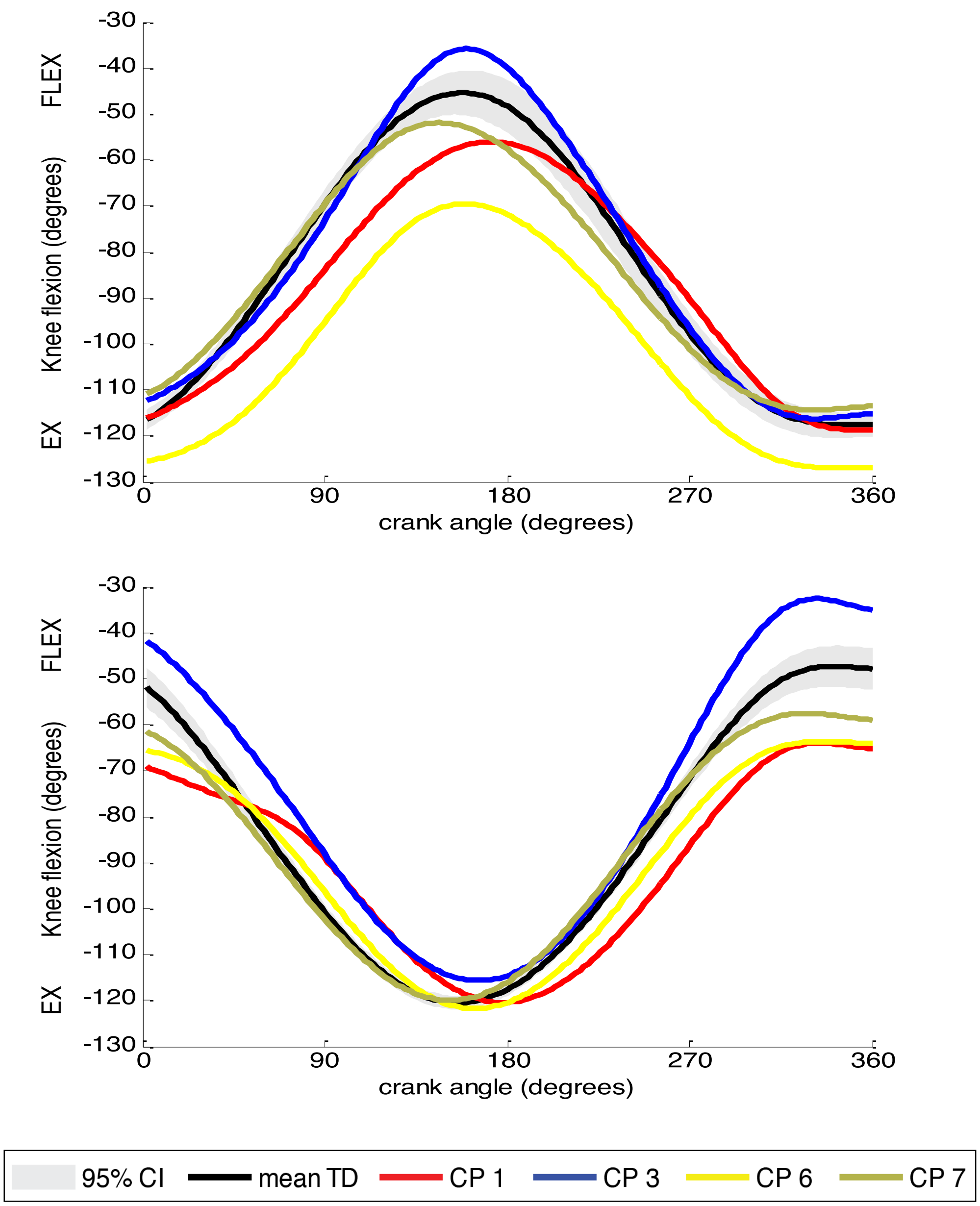


Figure A7: Knee flexion angles at 30 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION ANGLES

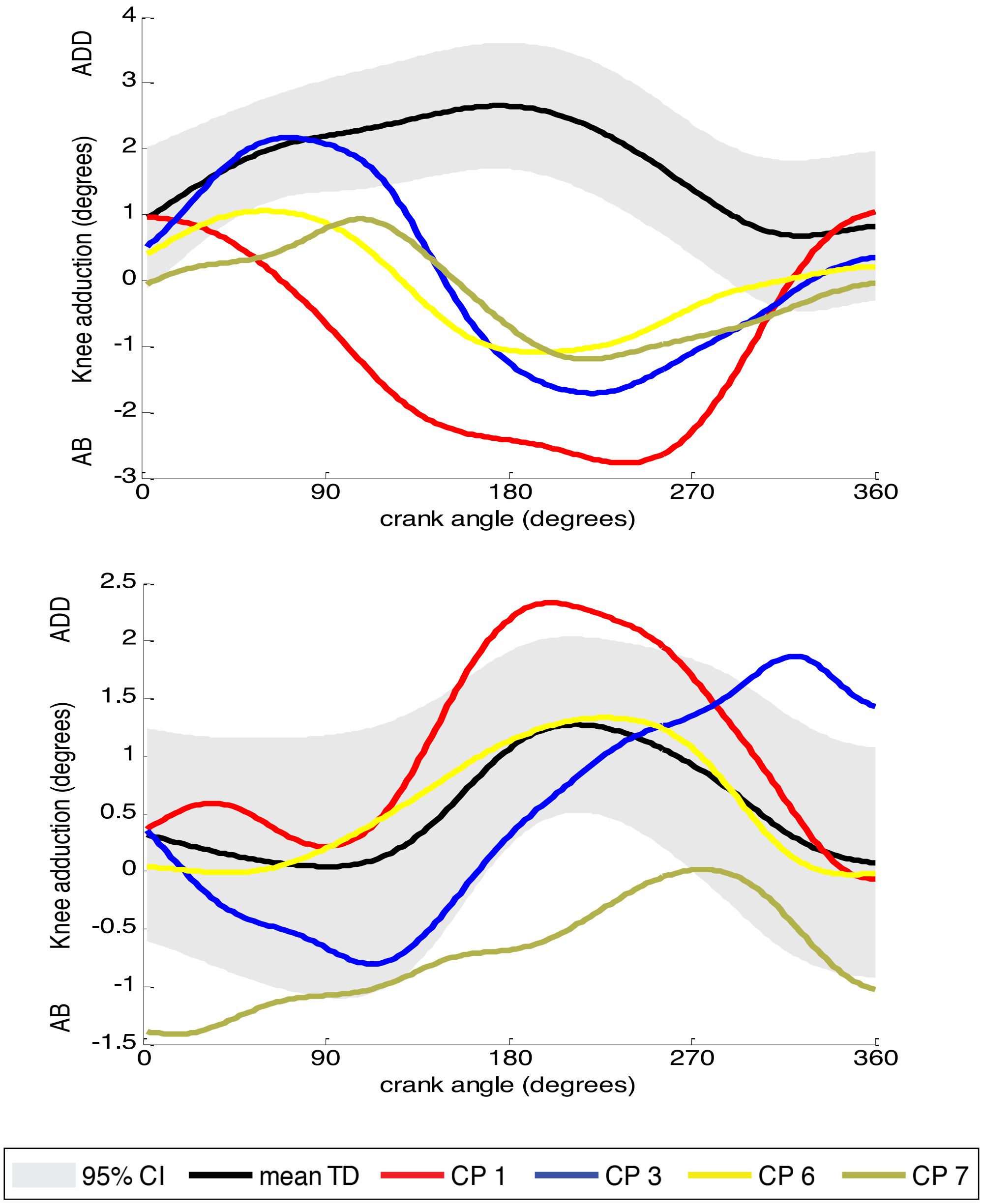


Figure A8: Knee adduction angles at 30 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION ANGLES

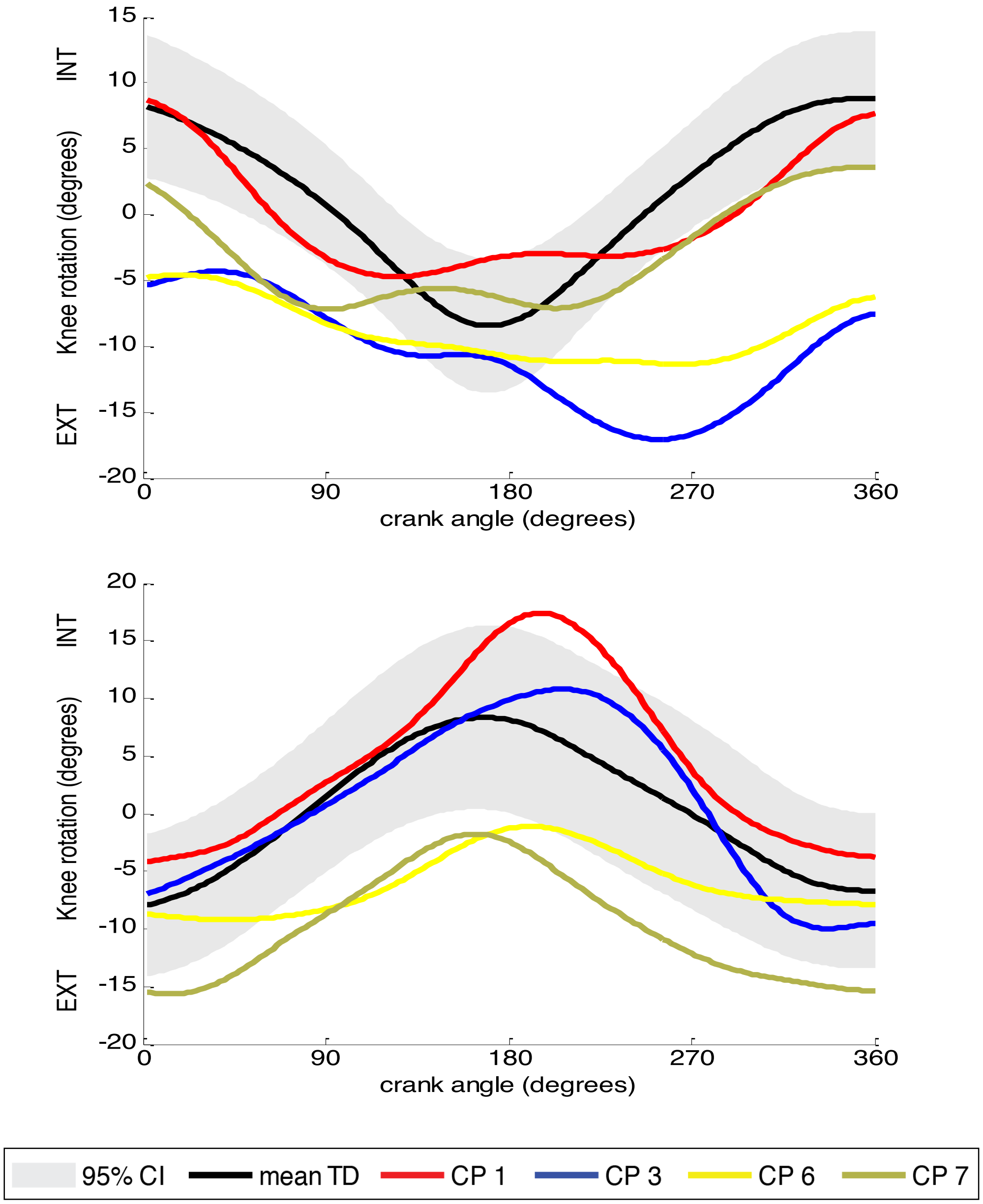


Figure A9: Knee rotation angles at 30 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION ANGLES

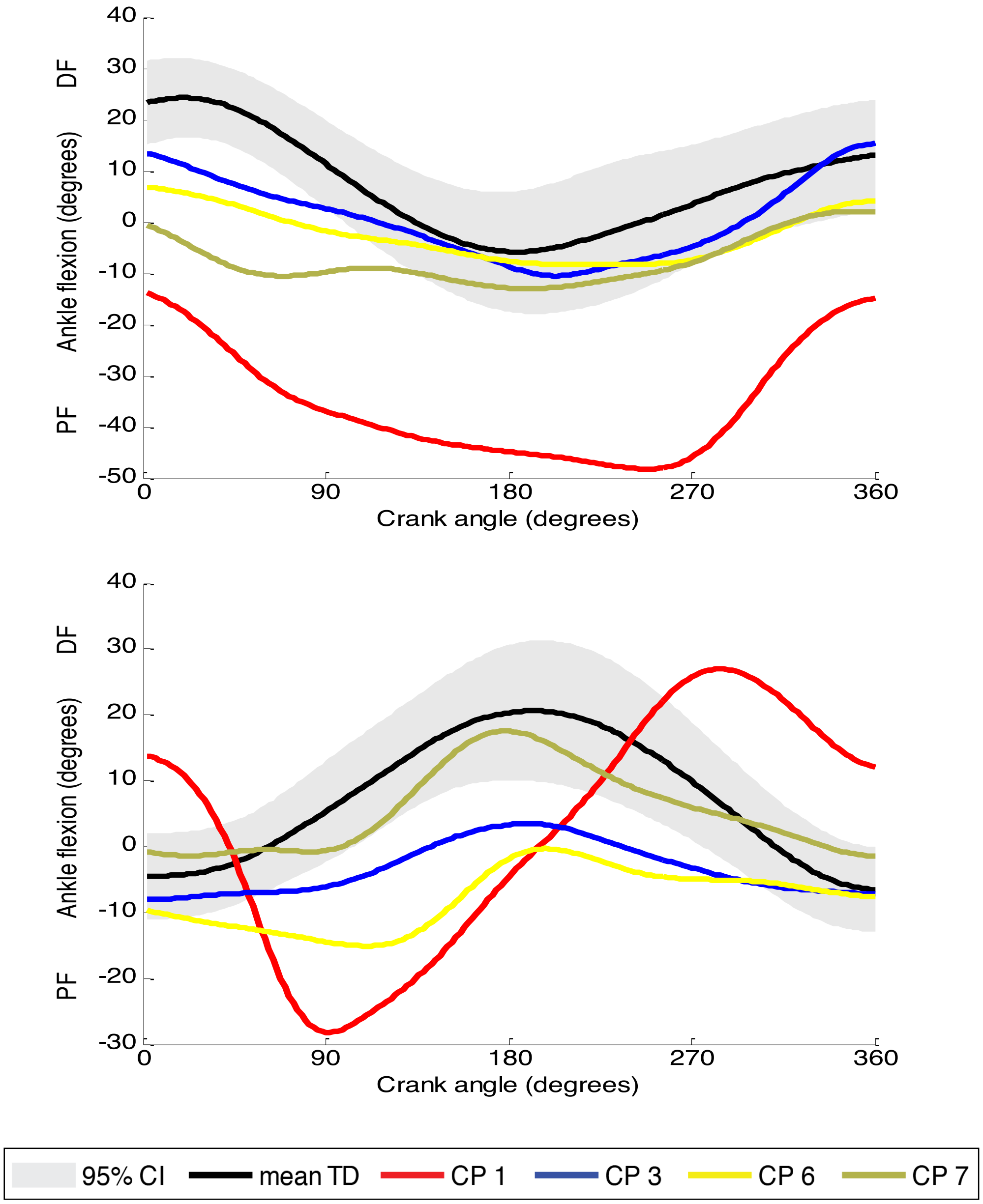


Figure A10: Ankle flexion angles at 30 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION ANGLES

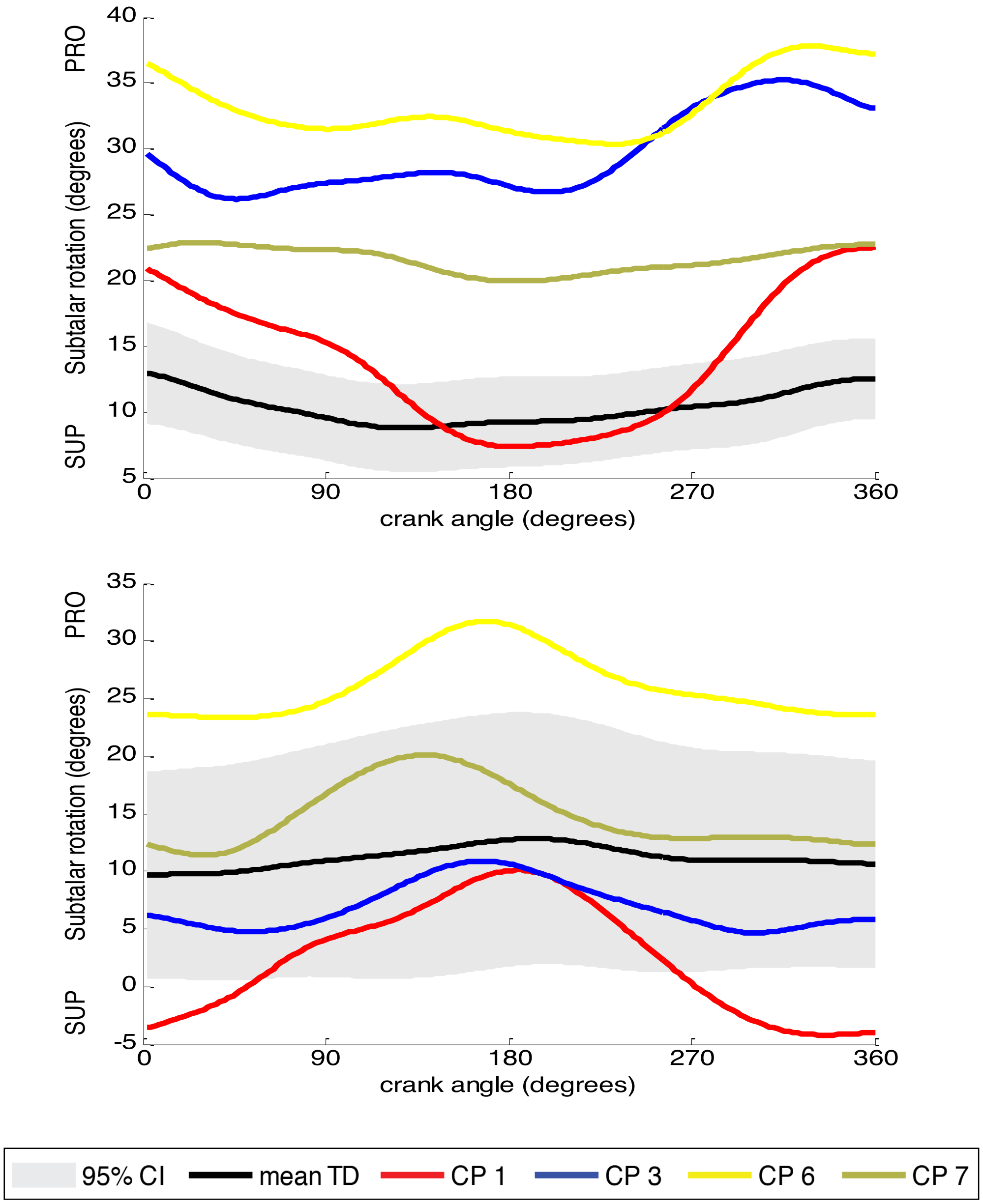


Figure A11: Subtalar rotation angles at 30 RPM produced by right (top) and left (bottom) leg.

APPENDIX M: JOINT ANGLES AT 40 RPM IN STUDY 2

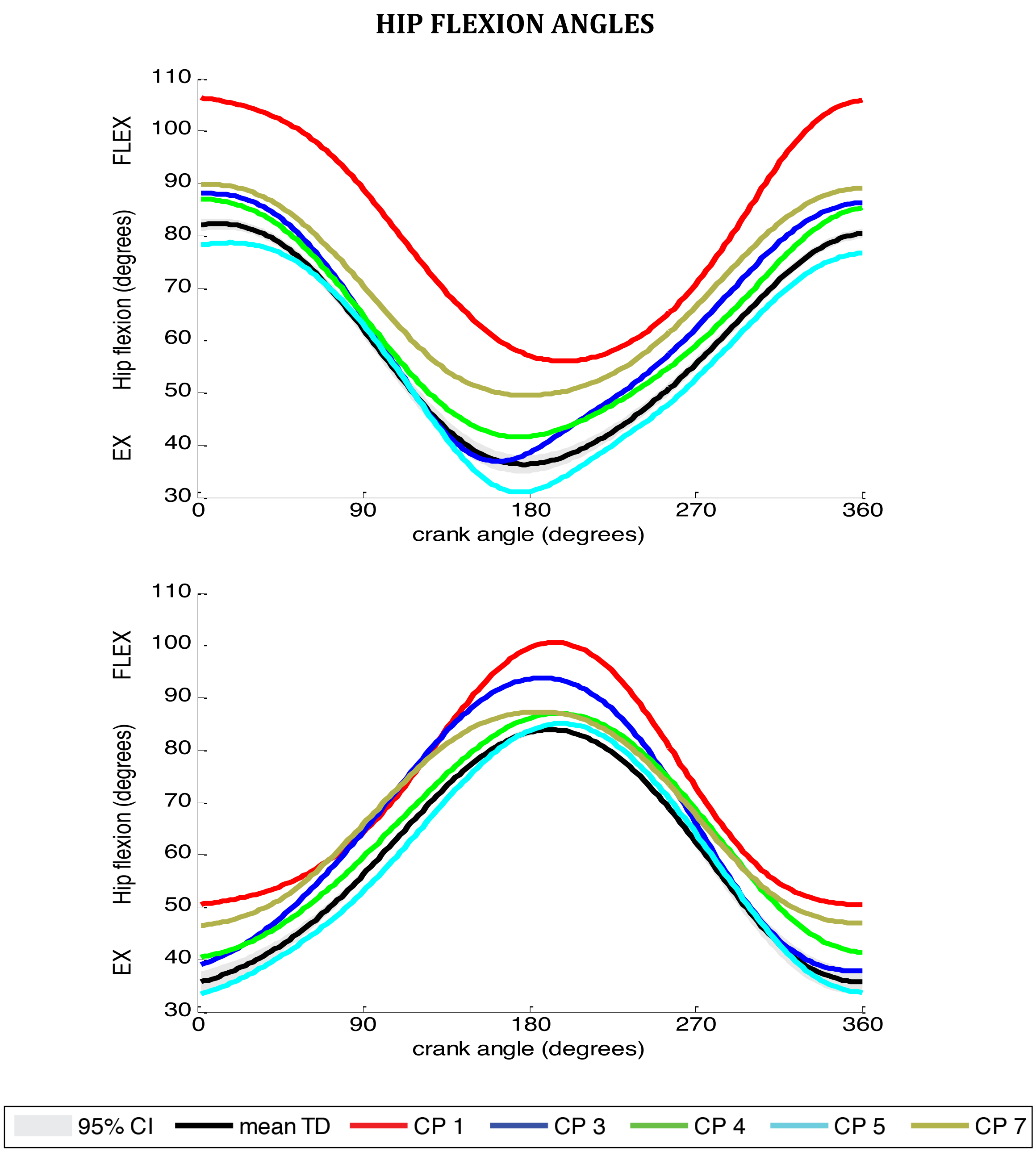


Figure A12: Hip flexion angles at 40 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION ANGLES

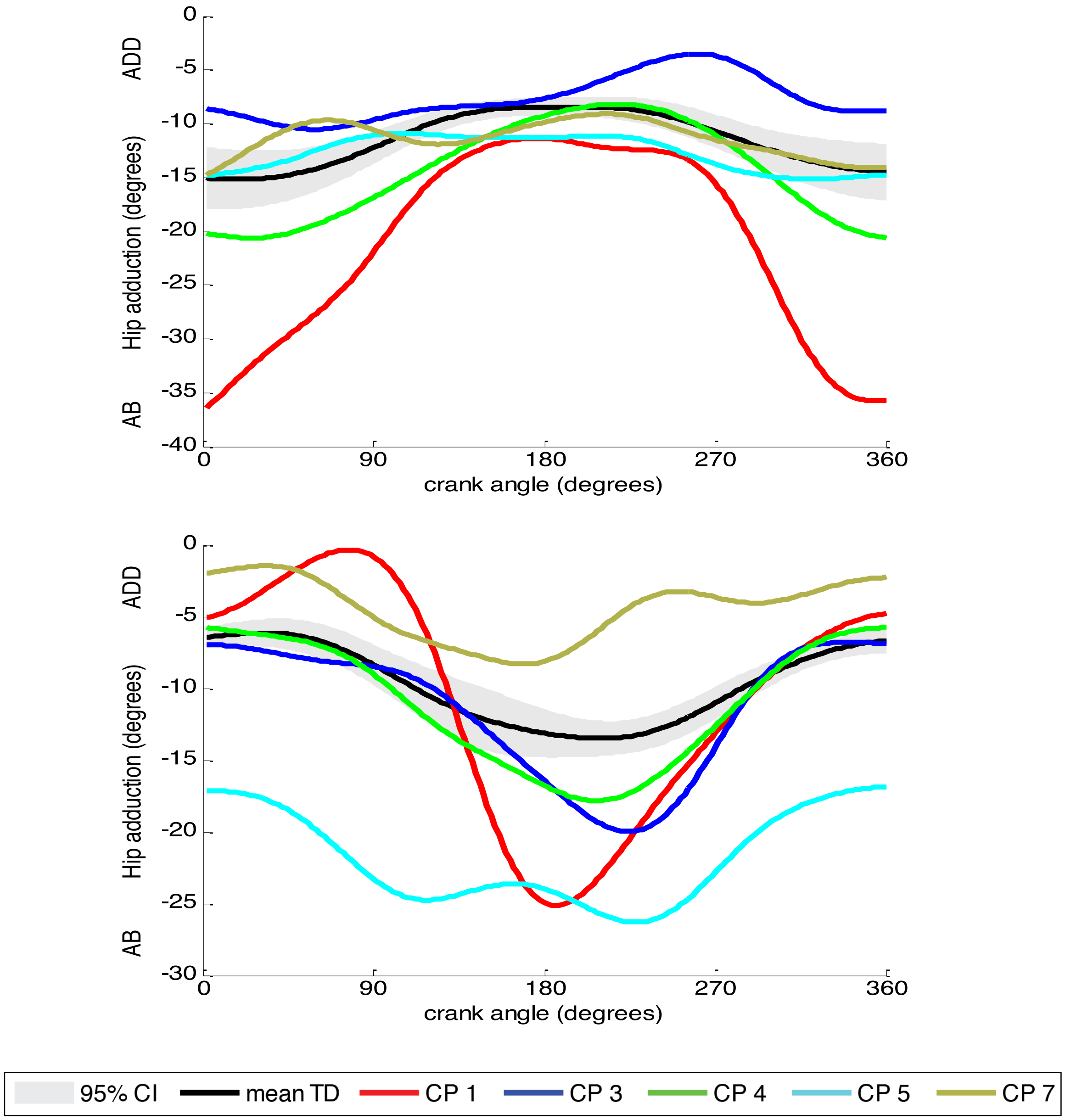


Figure A13: Hip adduction angles at 40 RPM produced by right (top) and left (bottom) leg.

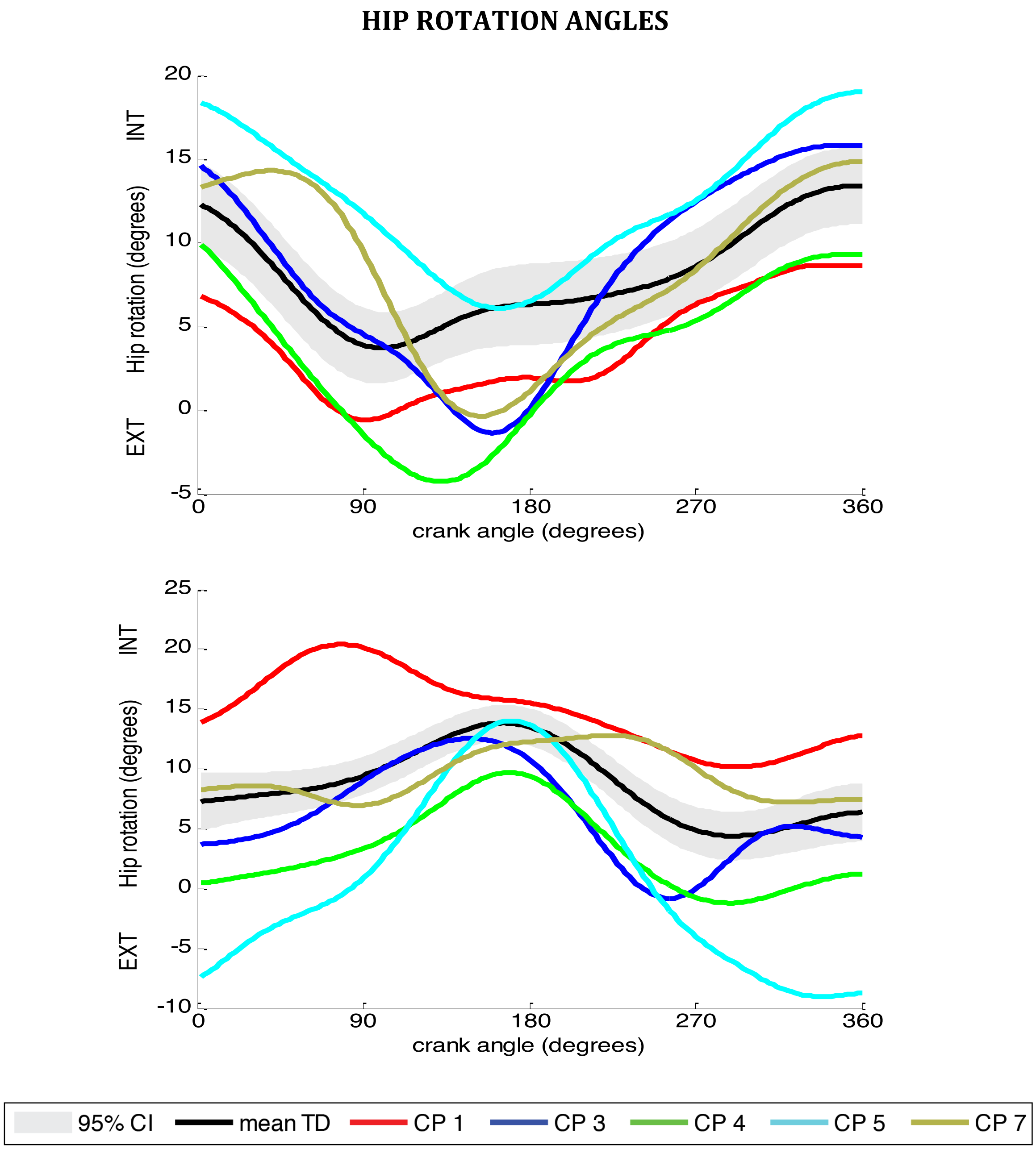


Figure A14: Hip rotation angles at 40 RPM produced by right (top) and left (bottom) leg.

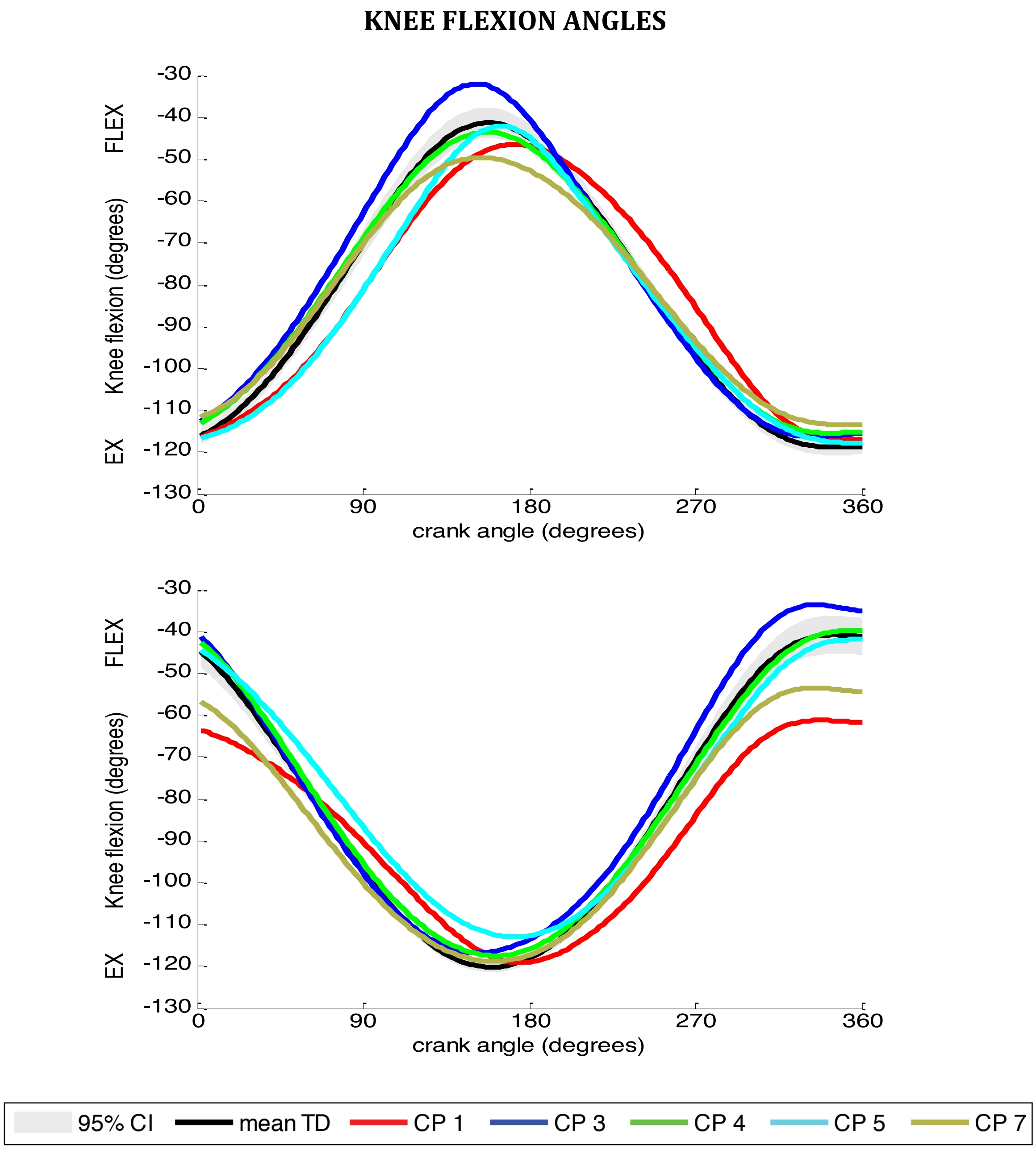


Figure A15: Knee flexion angles at 40 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION ANGLES

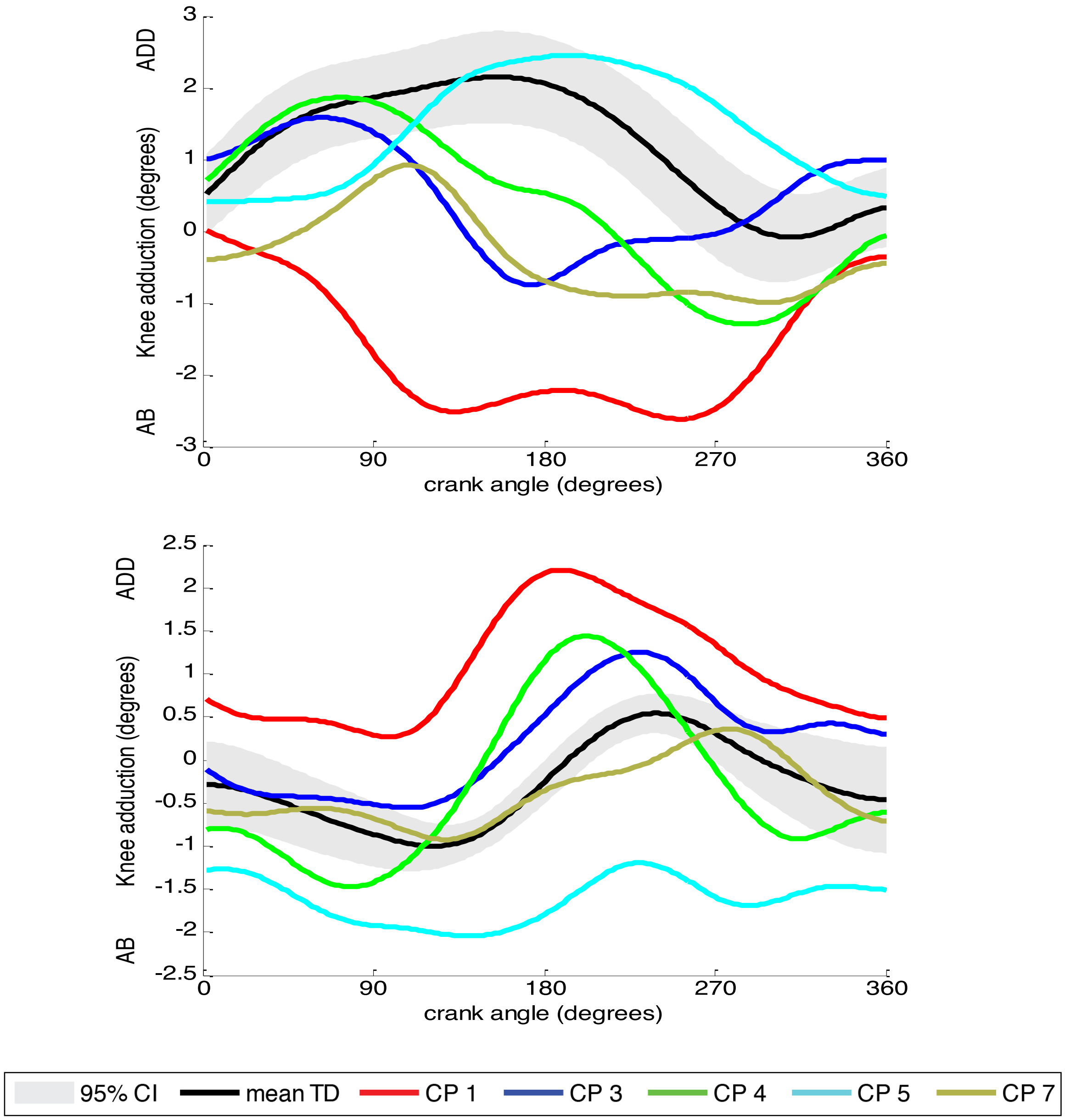


Figure A16: Knee adduction angles at 40 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION ANGLES

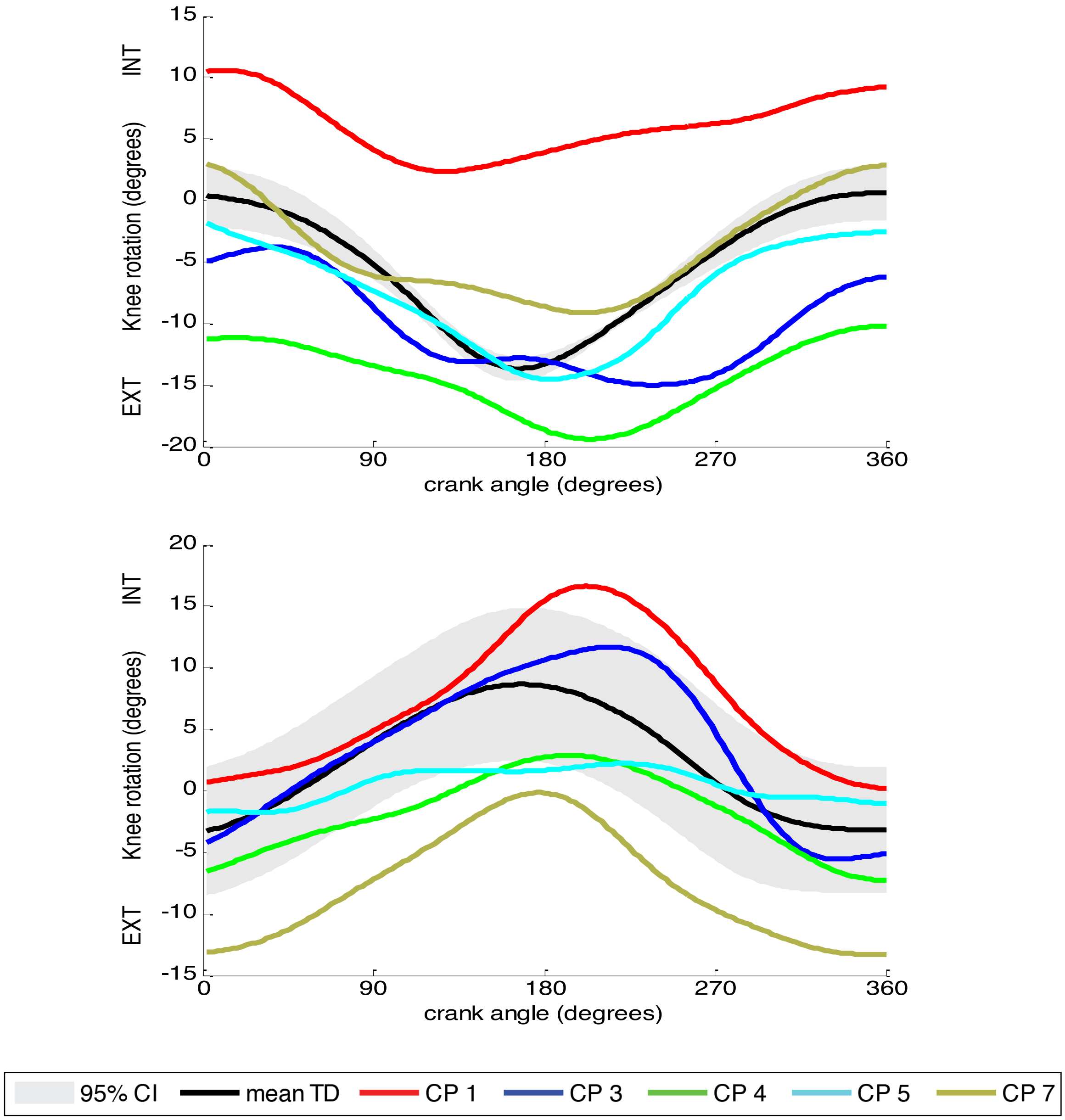
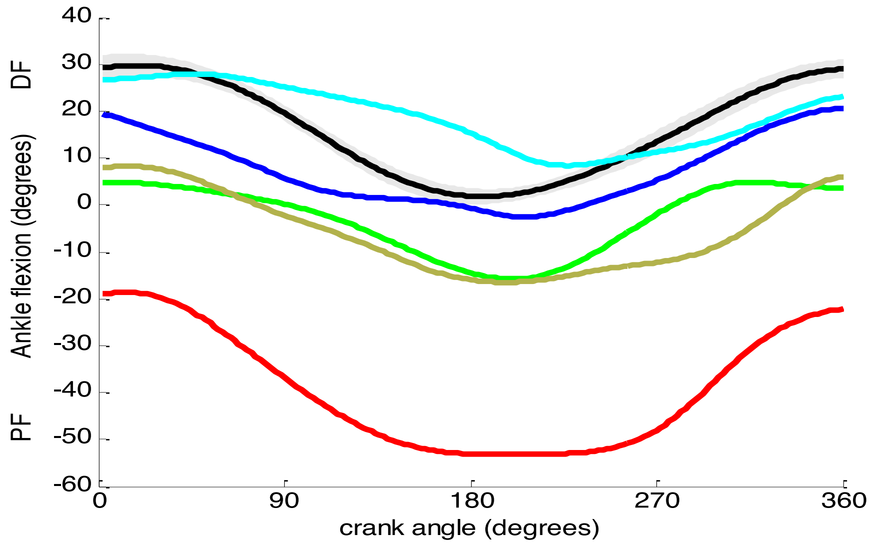


Figure A17: Knee rotation angles at 40 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION ANGLES



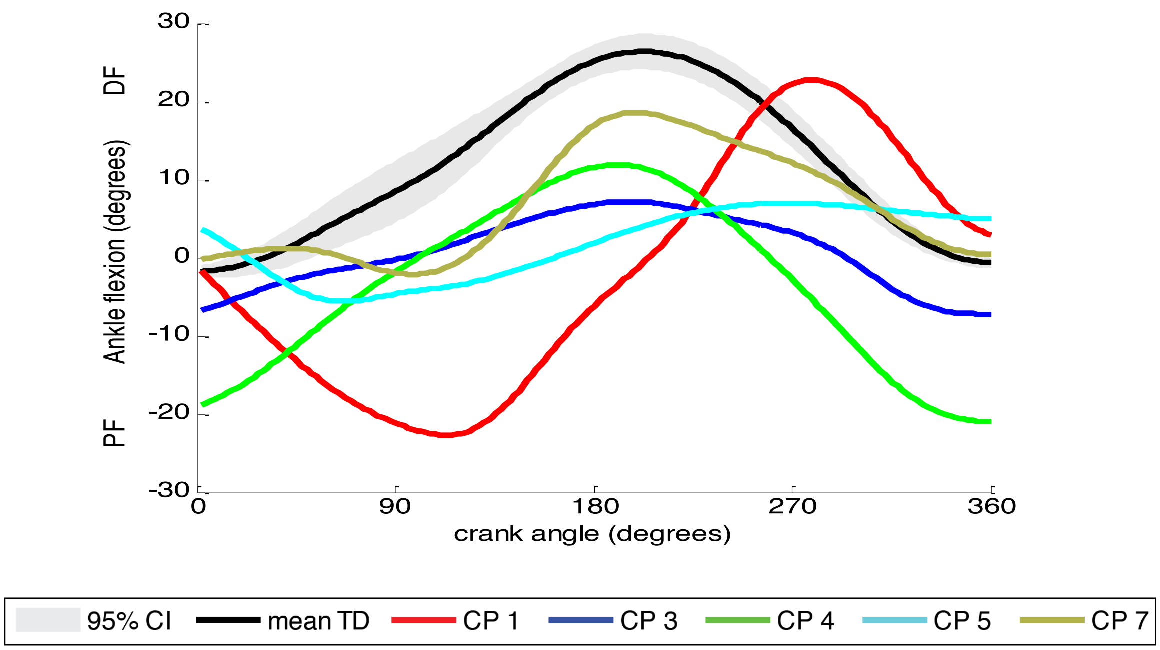


Figure A18: Ankle flexion angles at 40 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION ANGLES

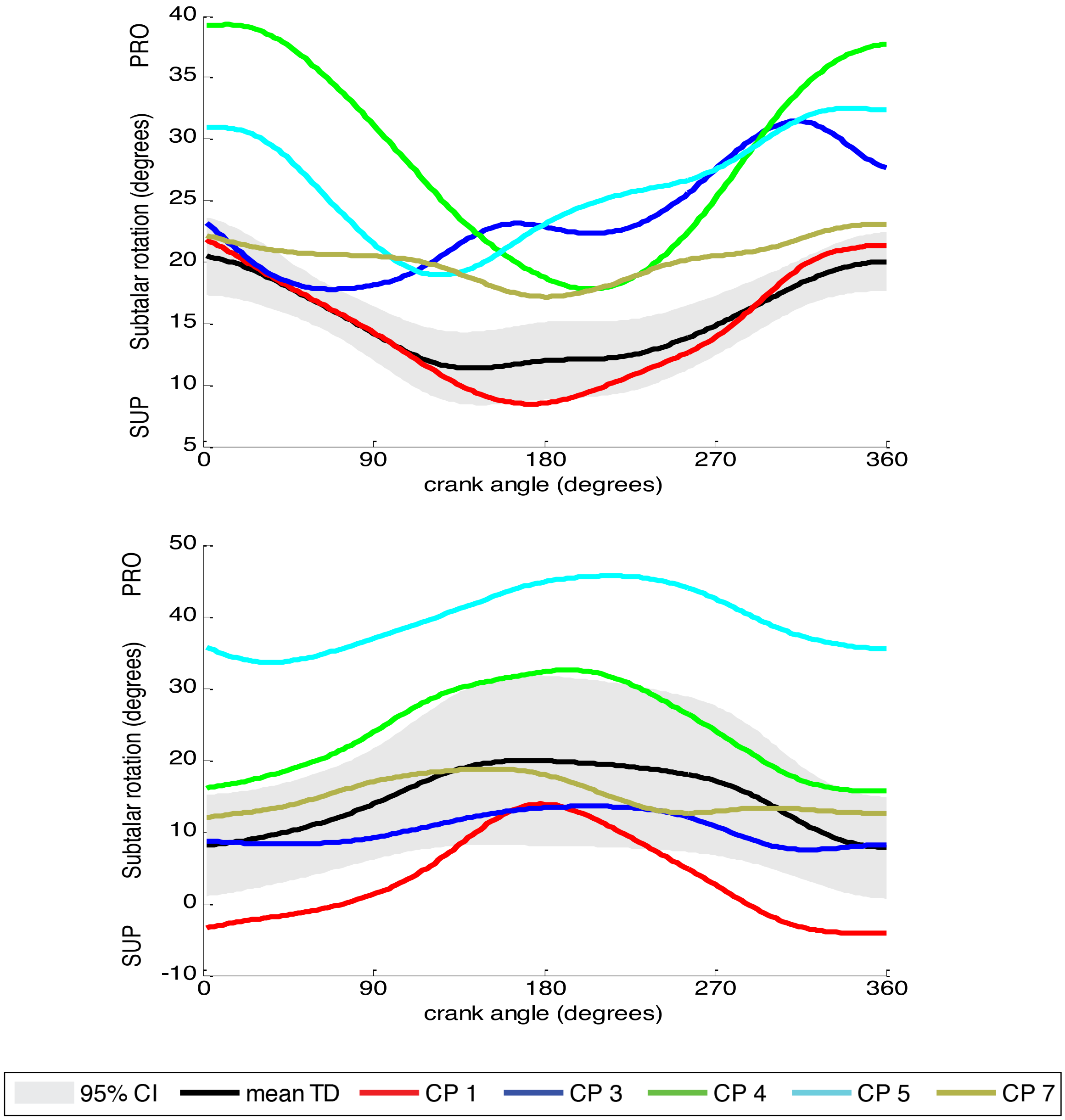


Figure A19: Subtalar rotation angles at 40 RPM produced by right (top) and left (bottom) leg.

APPENDIX N: JOINT ANGLES AT 50 RPM IN STUDY 2

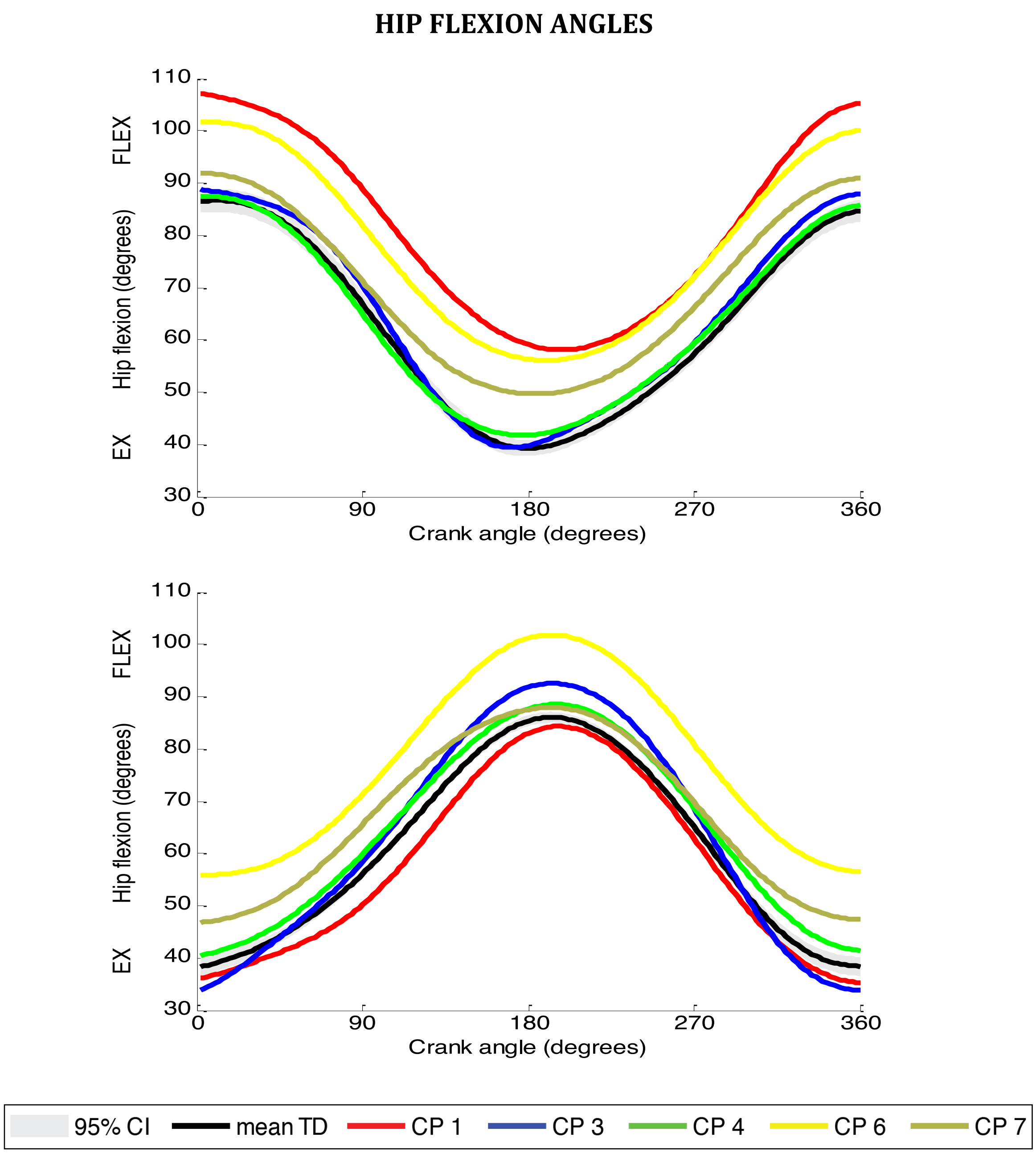


Figure A20: Hip flexion angles at 50 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION ANGLES

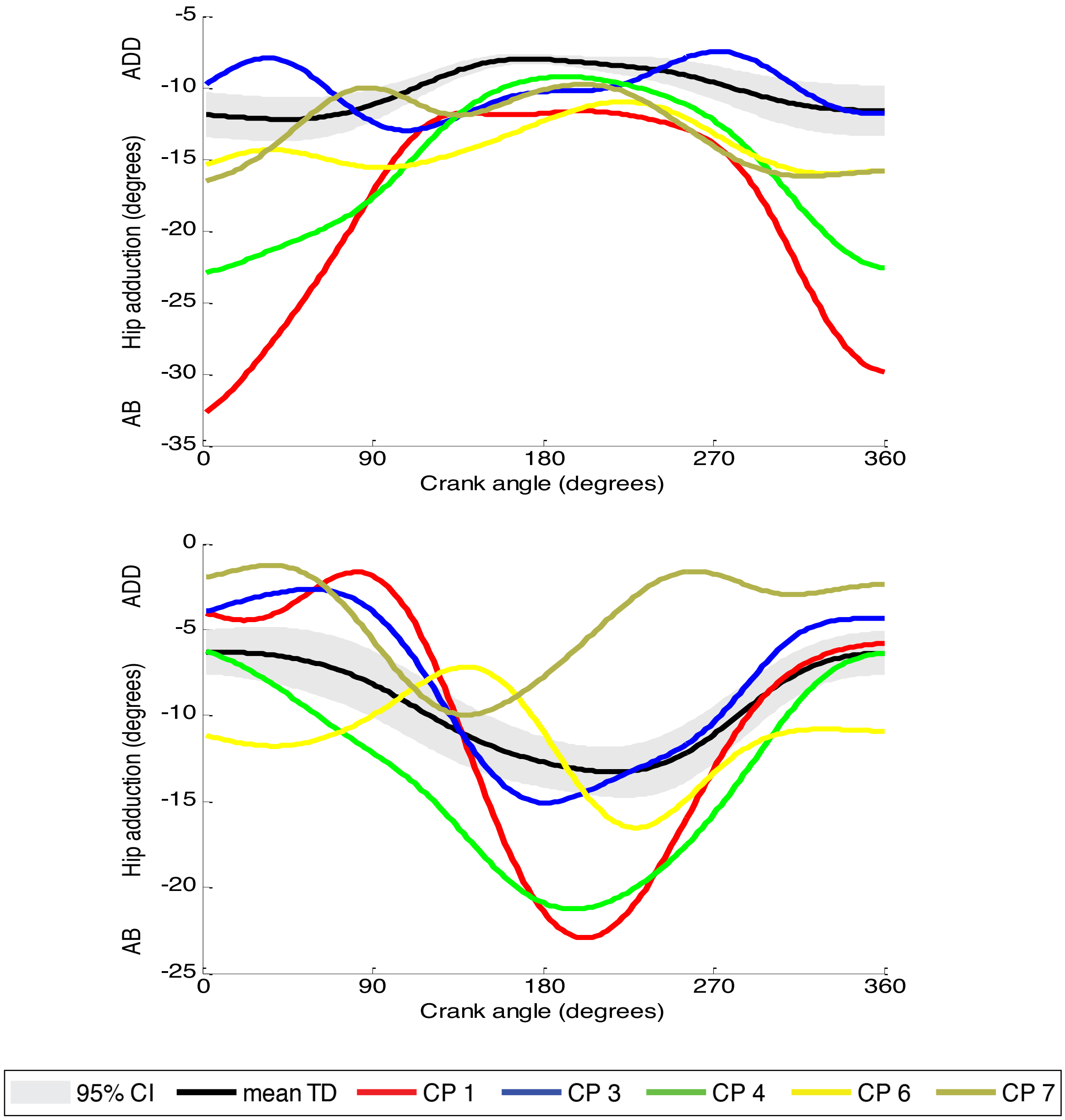


Figure A21: Hip adduction angles at 50 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION ANGLES

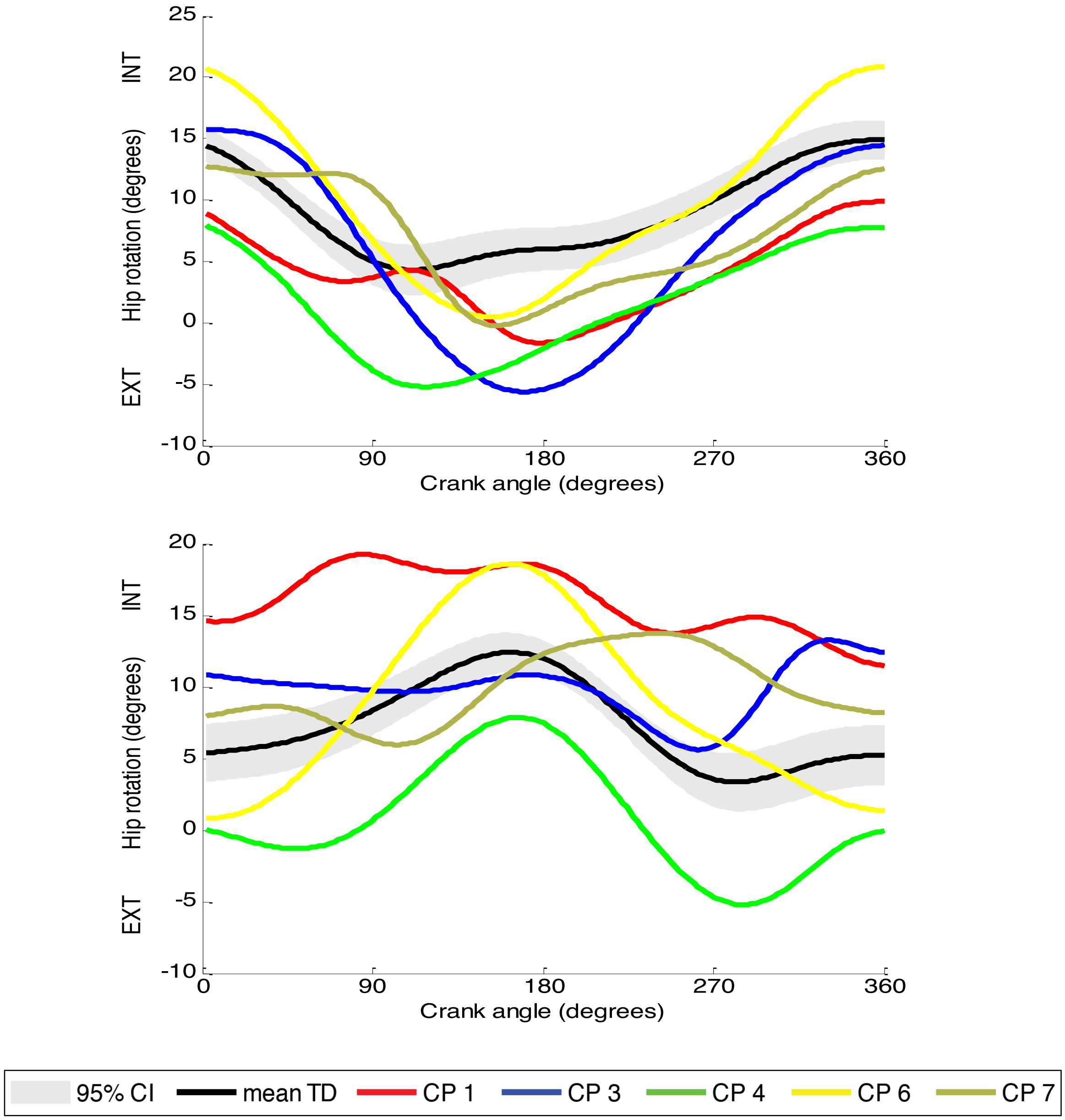


Figure A22: Hip rotation angles at 50 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION ANGLES

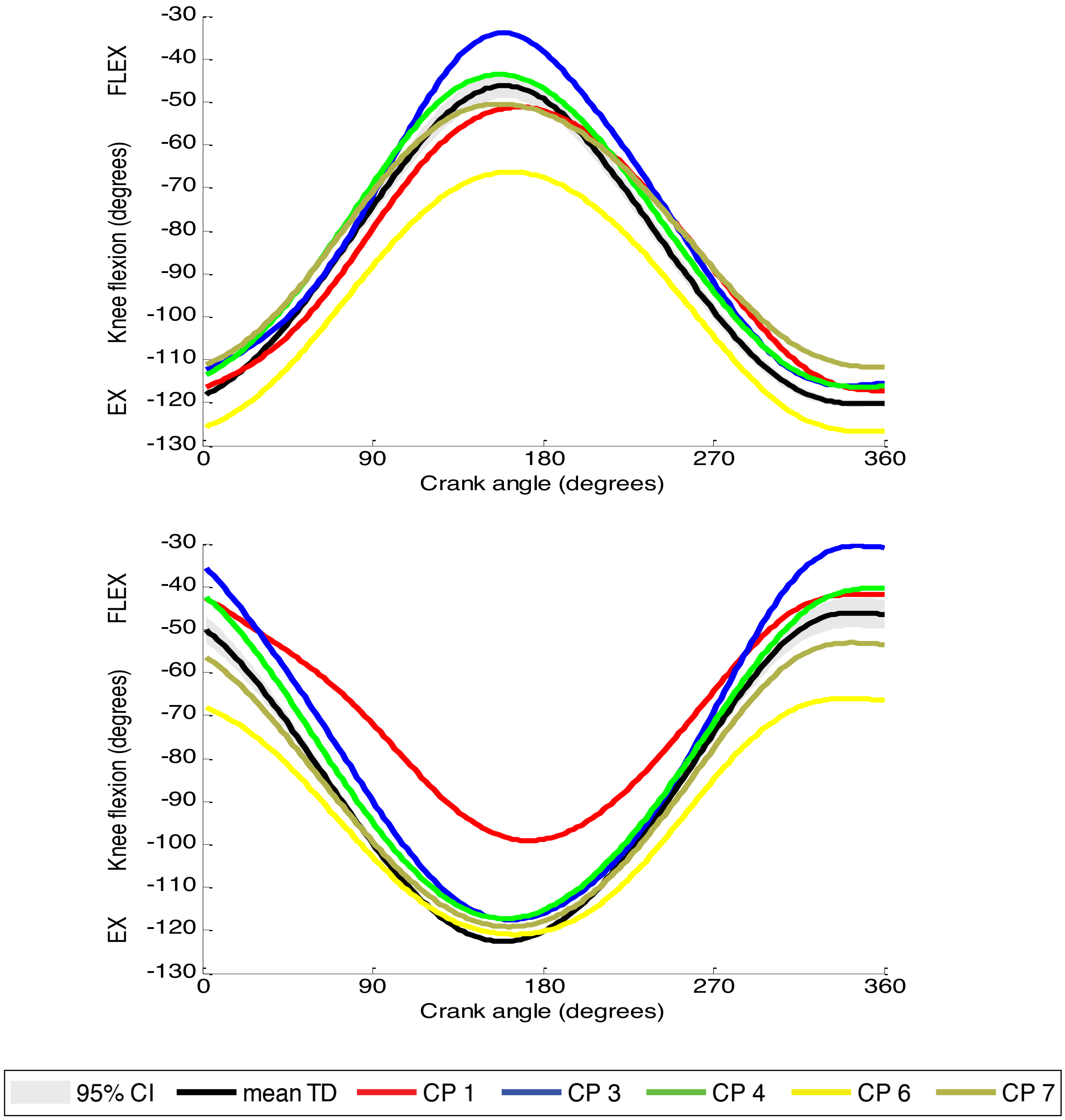


Figure A23: Knee flexion angles at 50 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION ANGLES

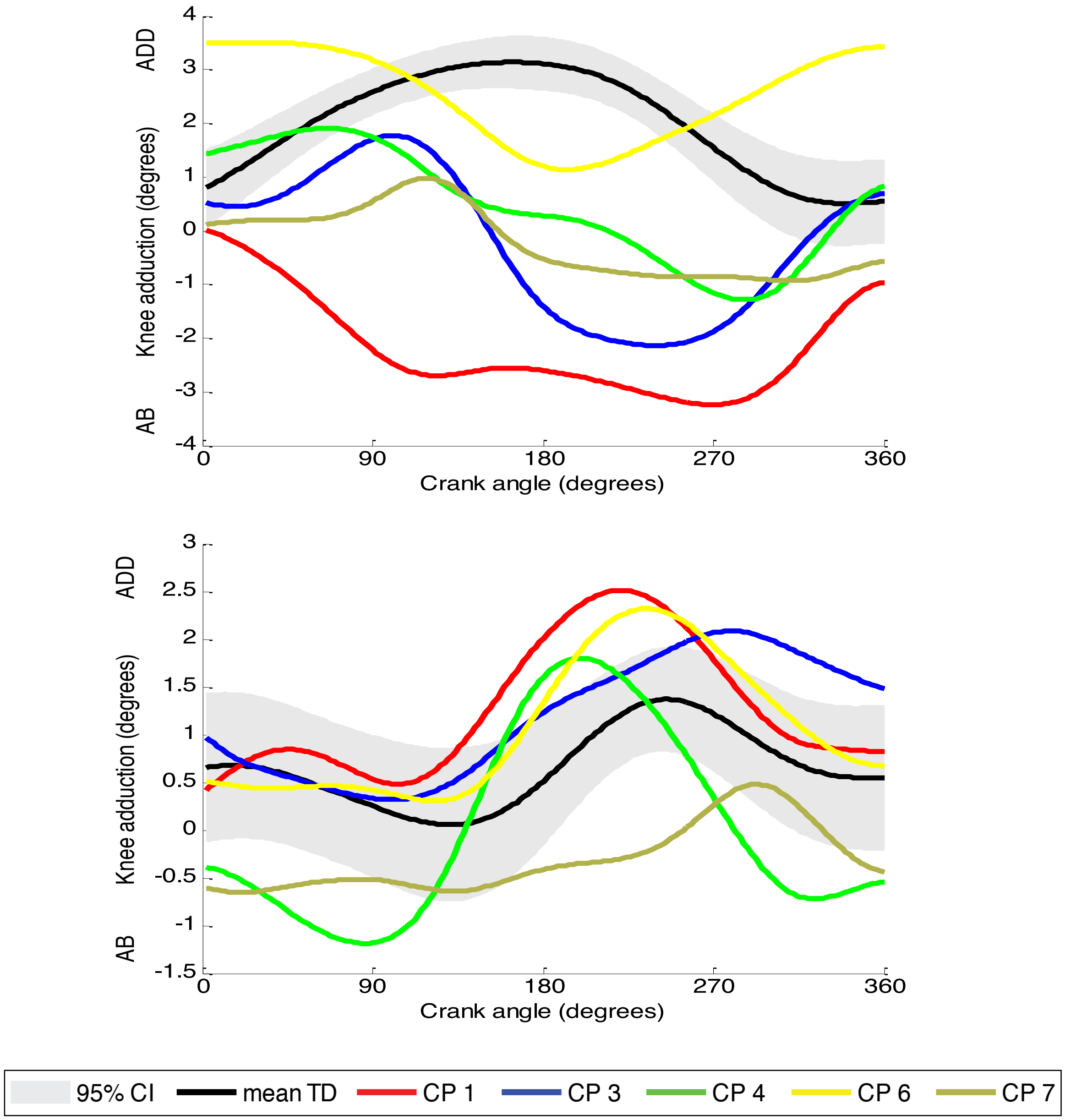


Figure A24: Knee adduction angles at 50 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION ANGLES

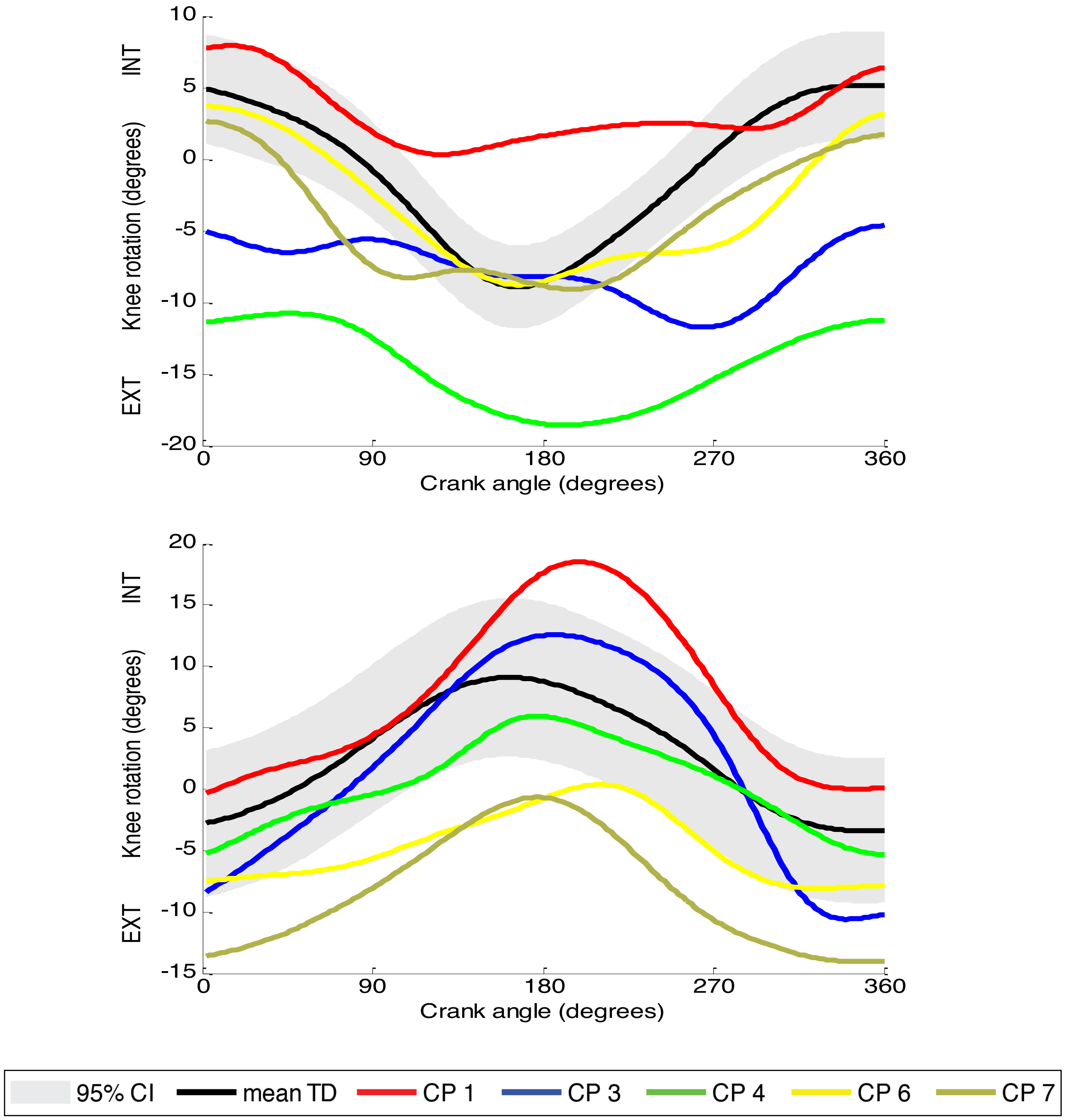


Figure A25: Knee rotation angles at 50 RPM produced by right (top) and left (bottom) leg.

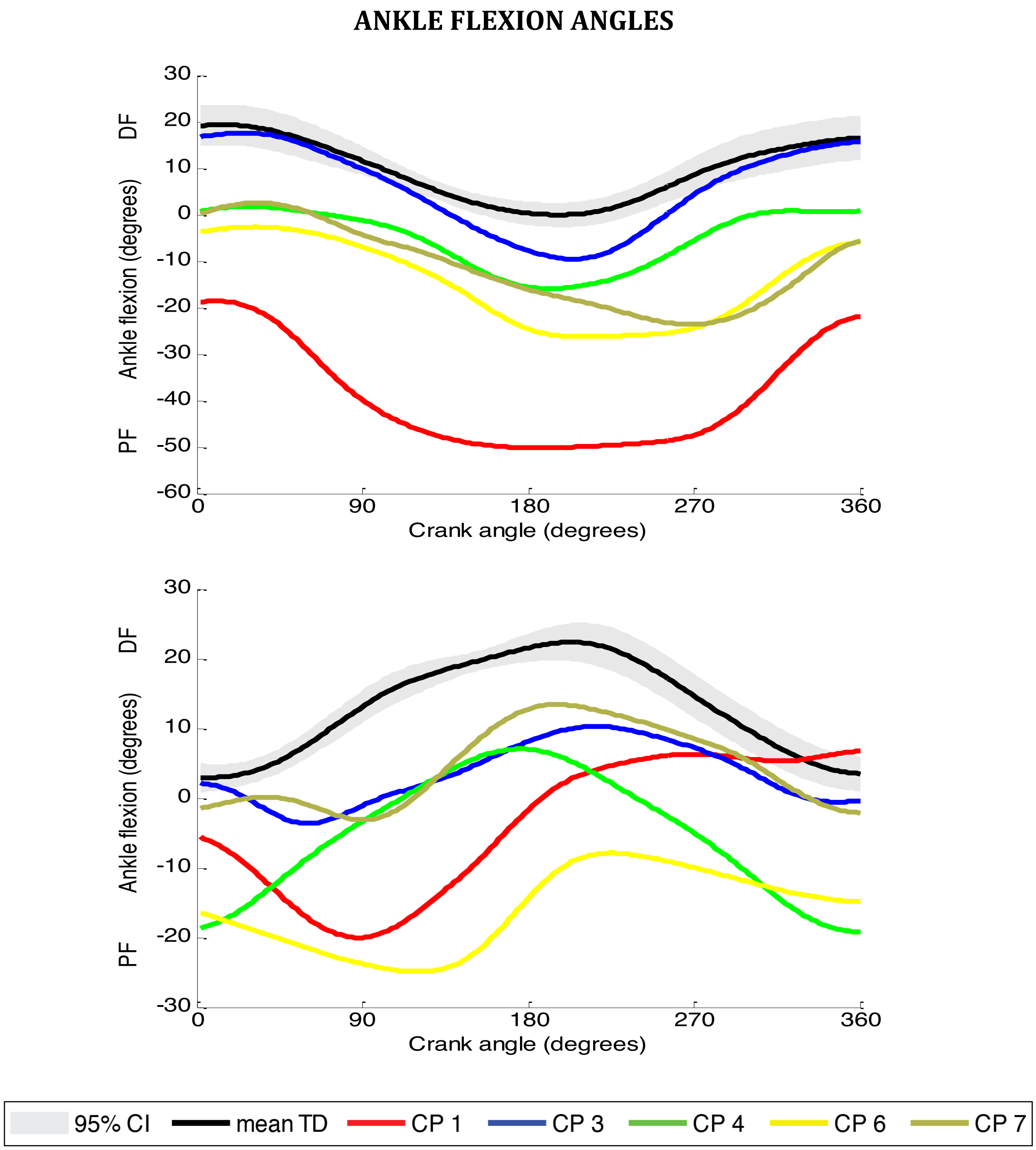


Figure A26: Ankle flexion angles at 50 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION ANGLES

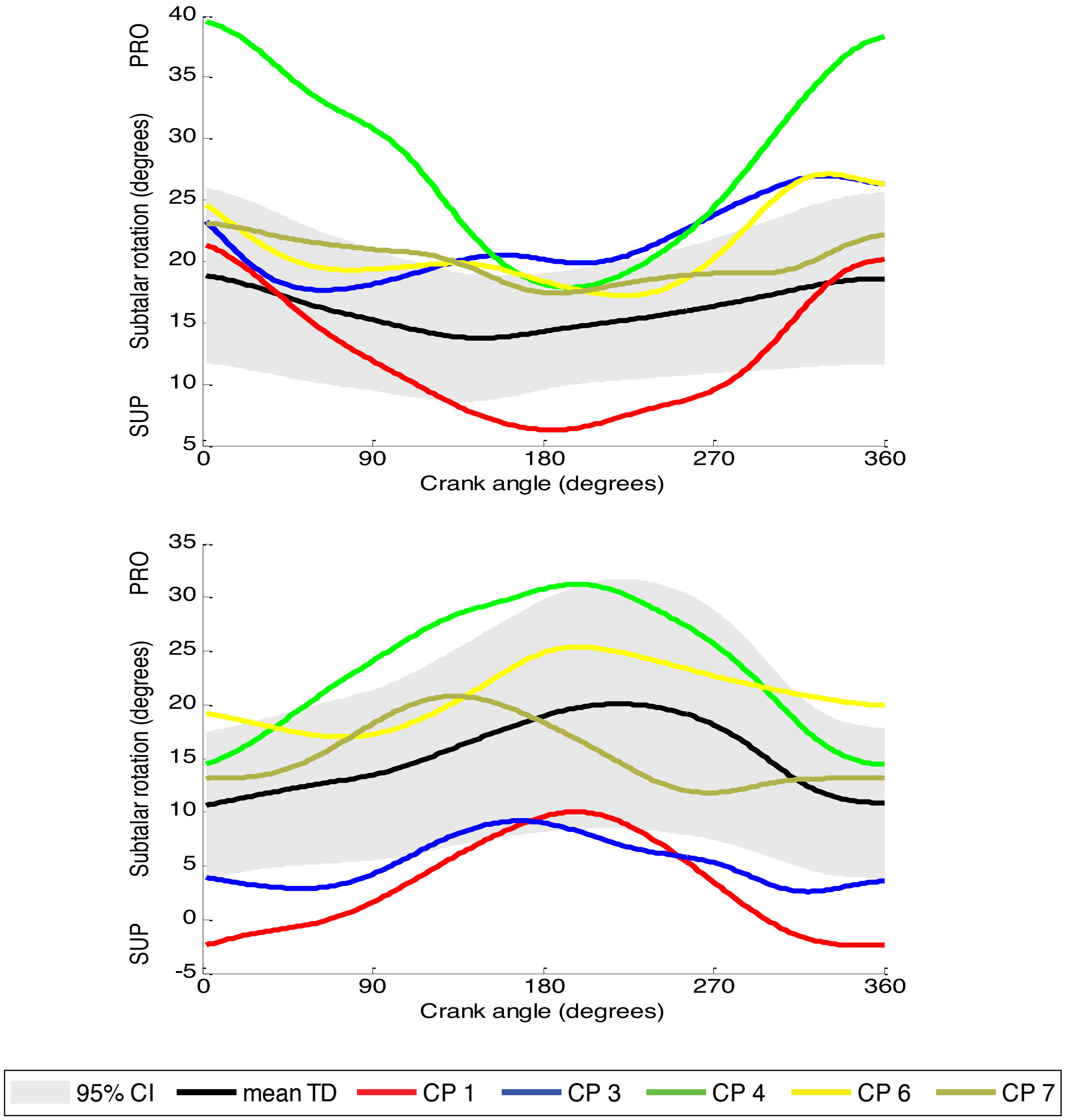


Figure A27: Subtalar rotation angles at 50 RPM produced by right (top) and left (bottom) leg.

APPENDIX O: JOINT ANGLES AT 60 RPM IN STUDY 2

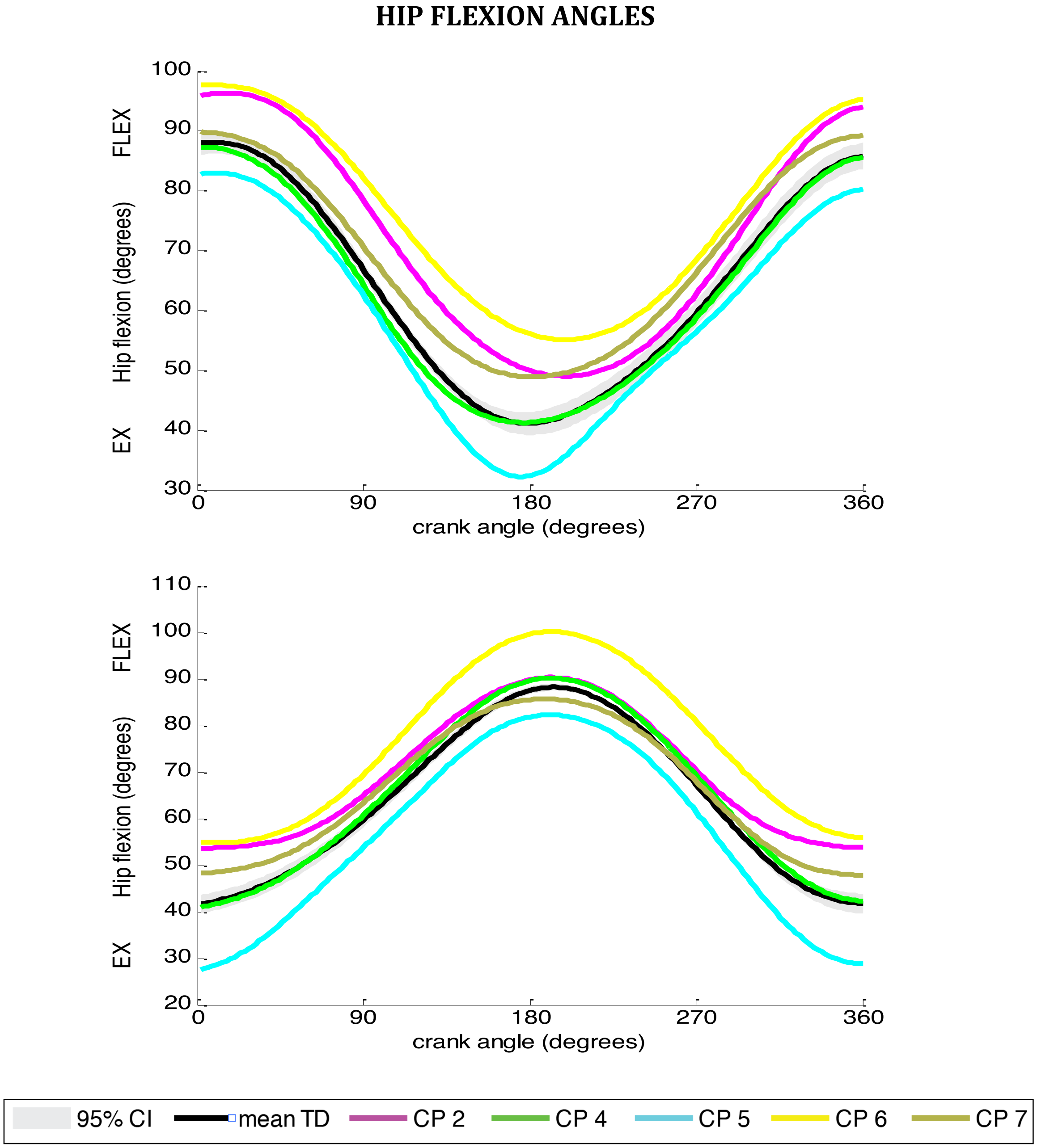


Figure A28: Hip flexion angles at 60 RPM produced by right (top) and left (bottom) leg.

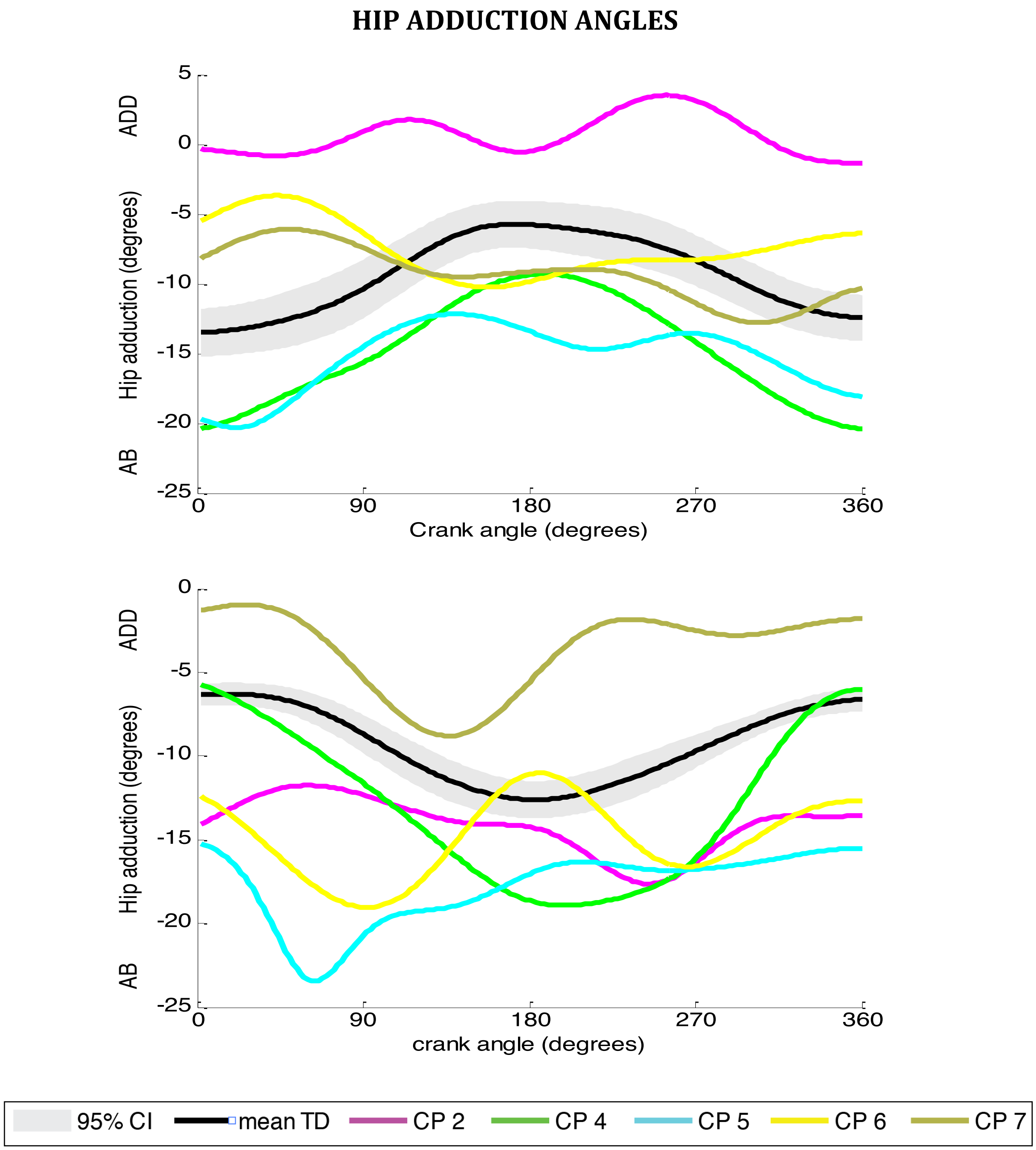


Figure A29: Hip adduction angles at 60 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION ANGLES

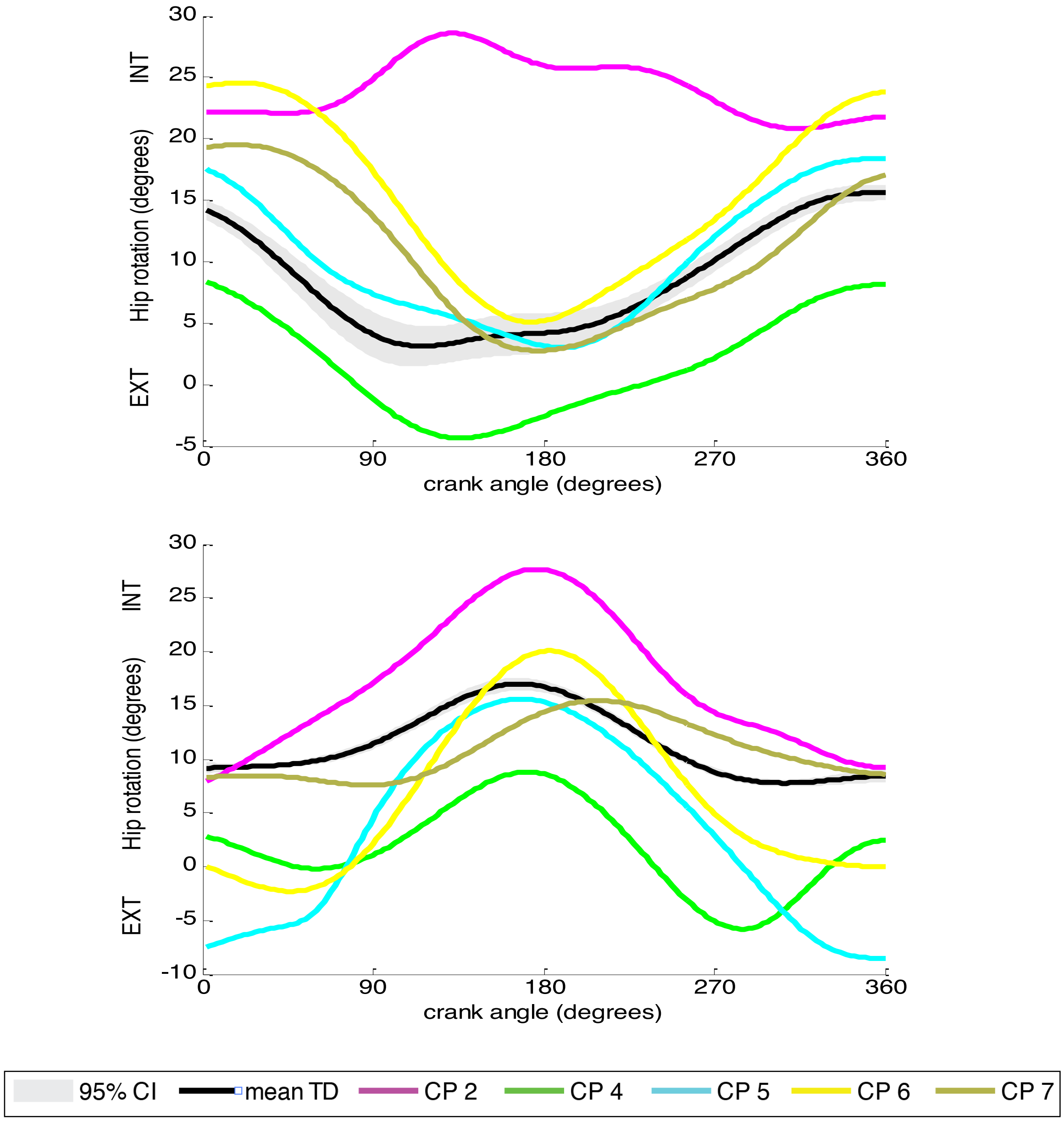


Figure A30: Hip rotation angles at 60 RPM produced by right (top) and left (bottom) leg.

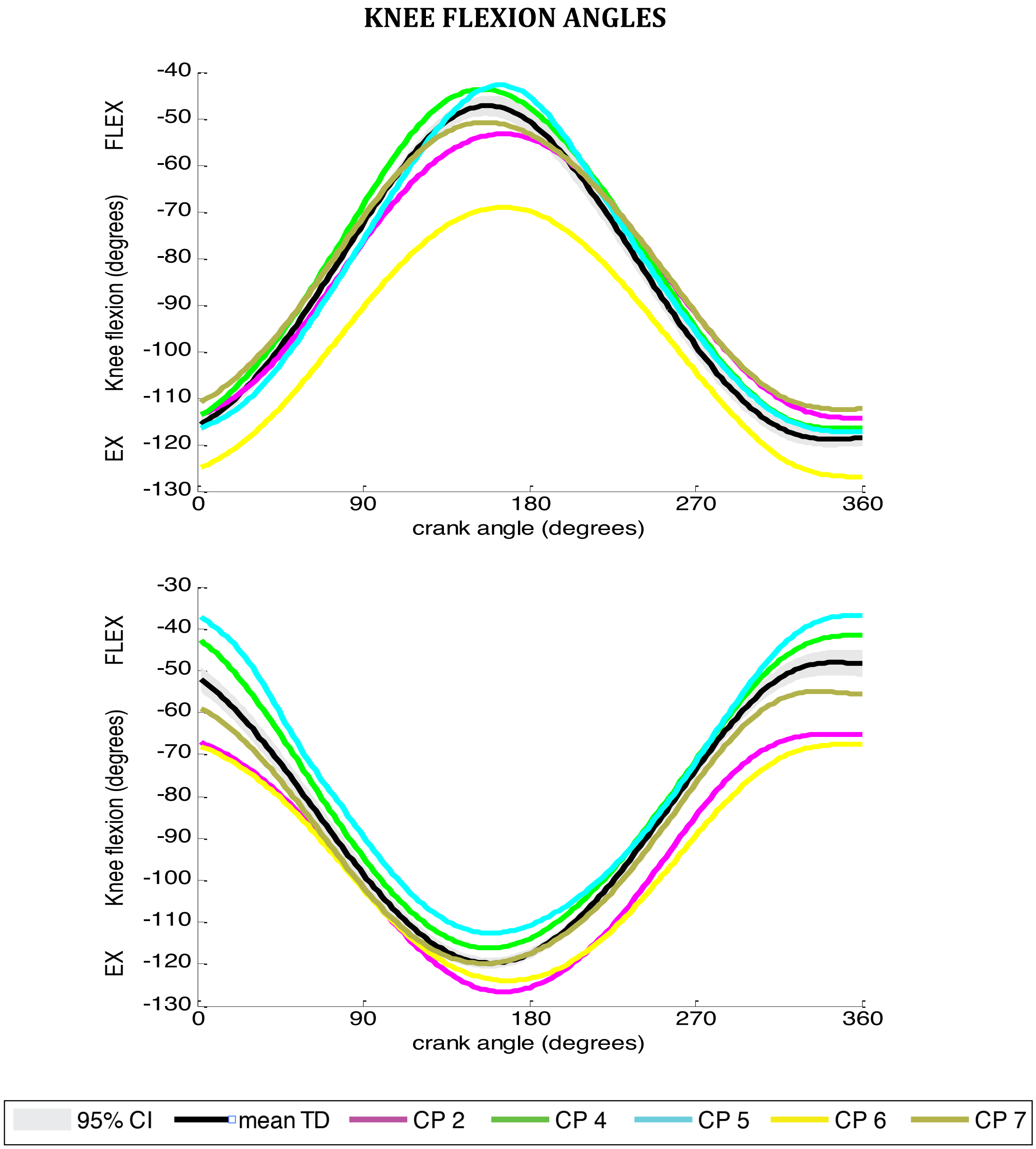


Figure A31: Knee flexion angles at 60 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION ANGLES

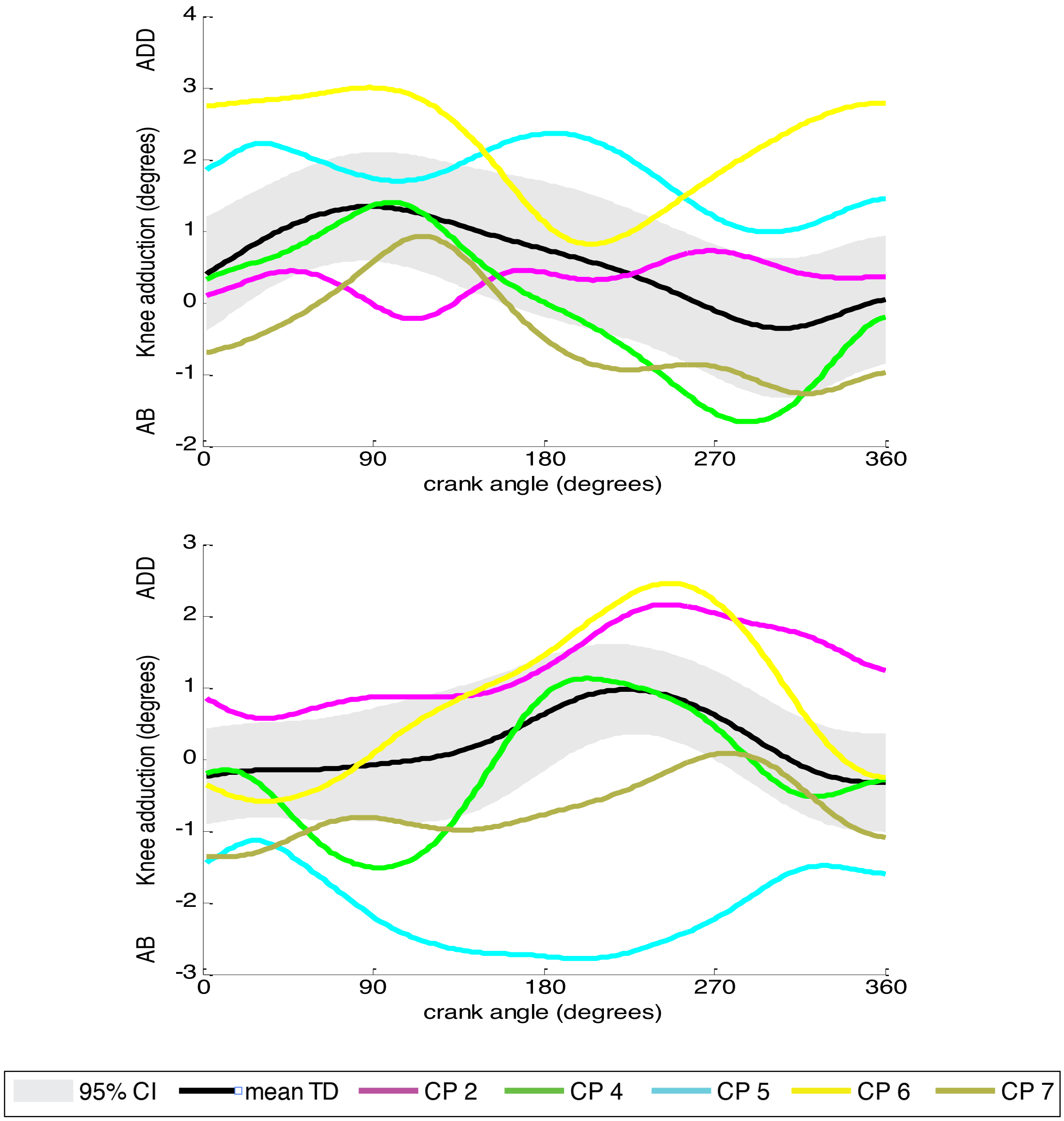


Figure A32: Knee adduction angles at 60 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION ANGLES

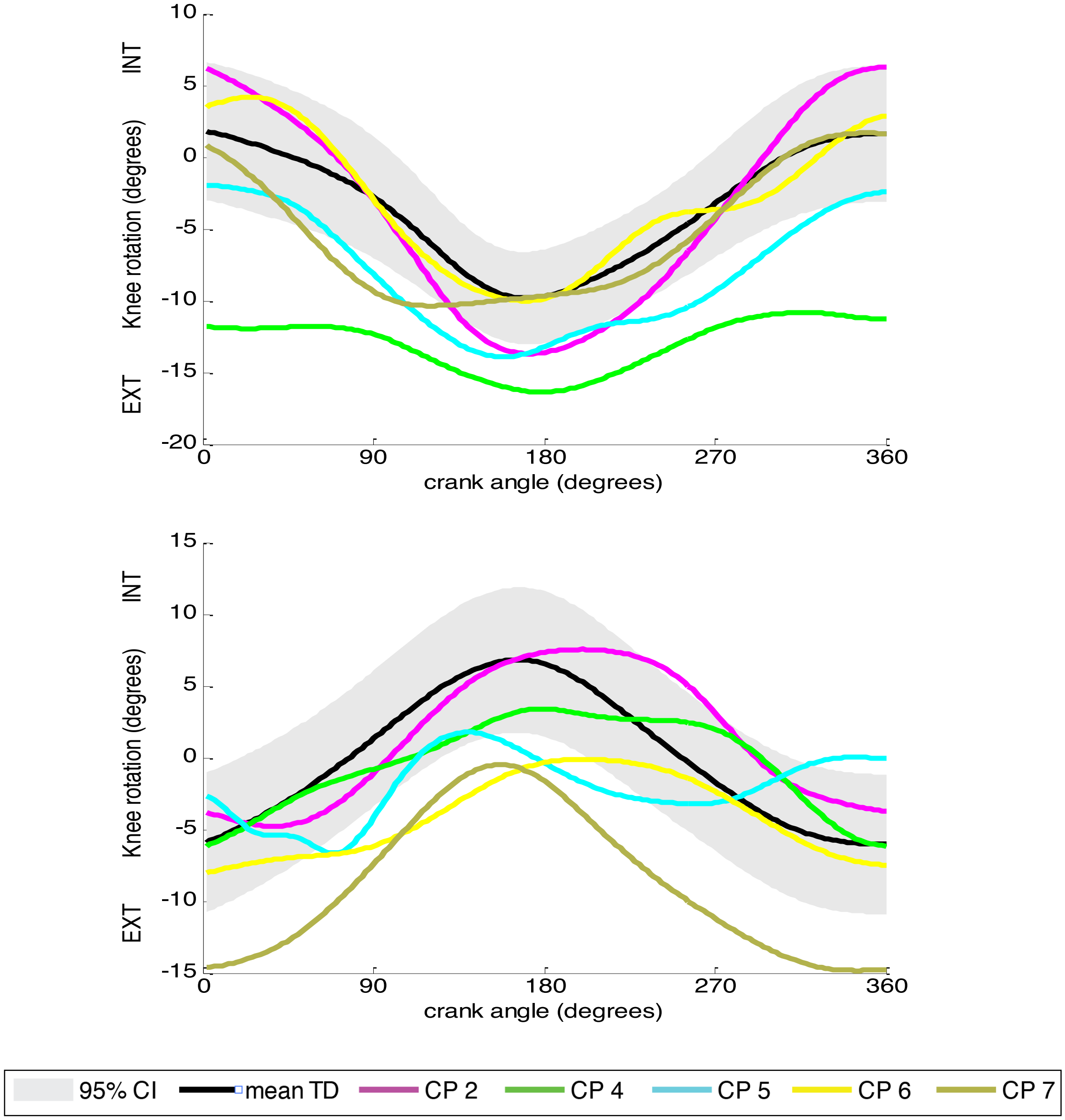


Figure A33: Knee rotation angles at 60 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION ANGLES

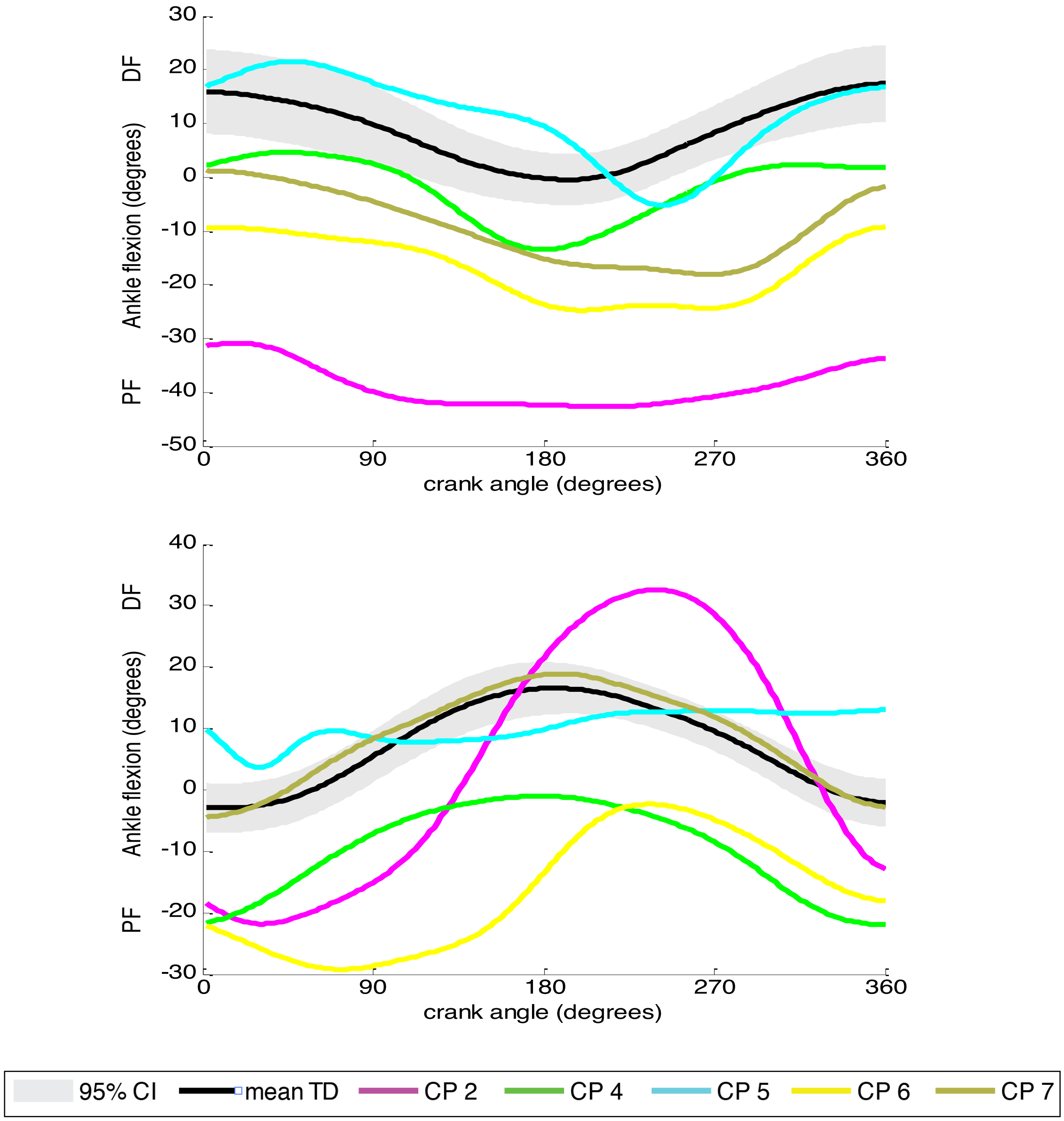


Figure A34: Ankle flexion angles at 60 RPM produced by right (top) and left (bottom) leg.

SUBTALAR FLEXION ANGLES

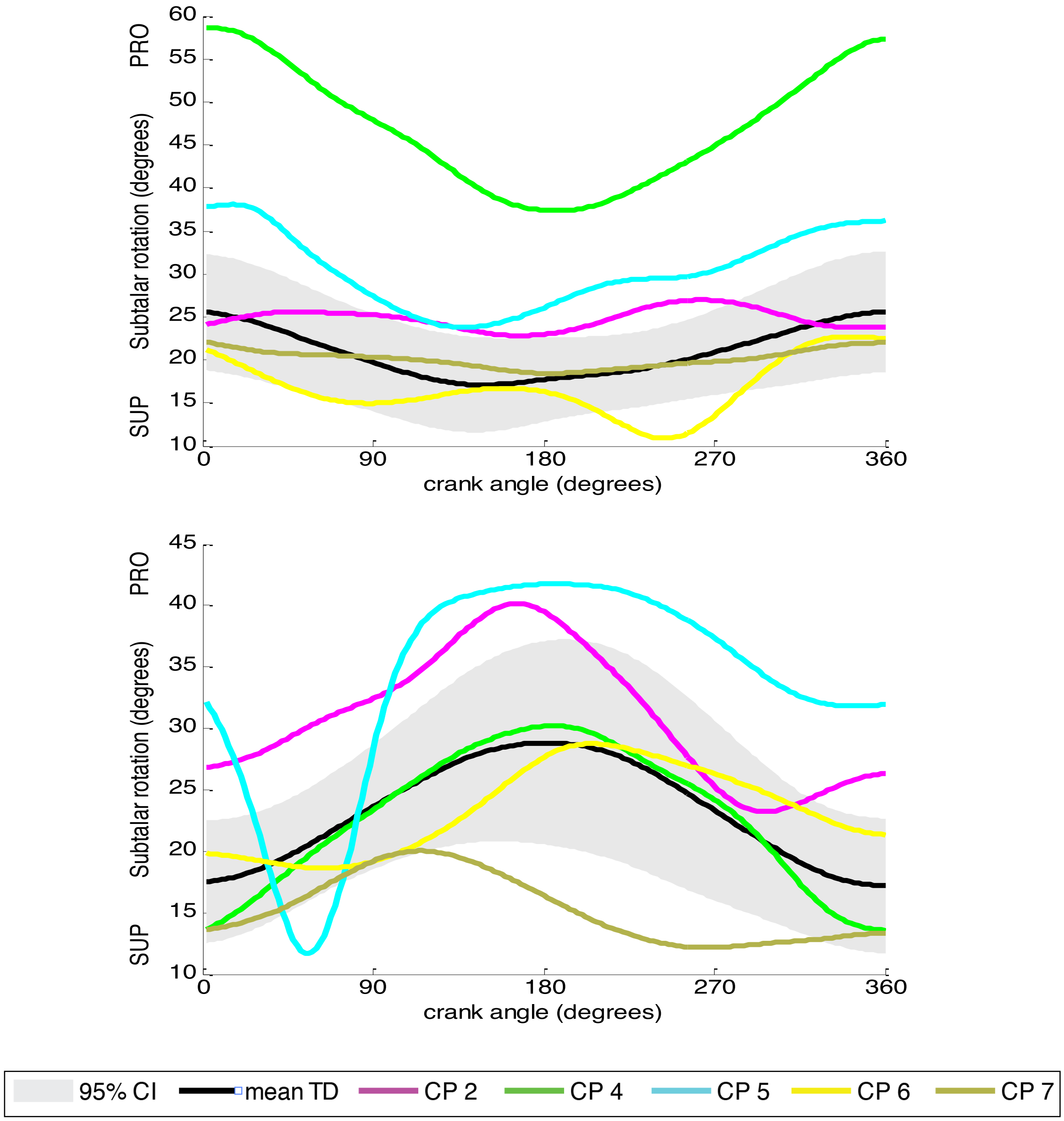


Figure A35: Subtalar rotation angles at 60 RPM produced by right (top) and left (bottom) leg.

APPENDIX P: JOINT ANGLES AT 70 RPM IN STUDY 2

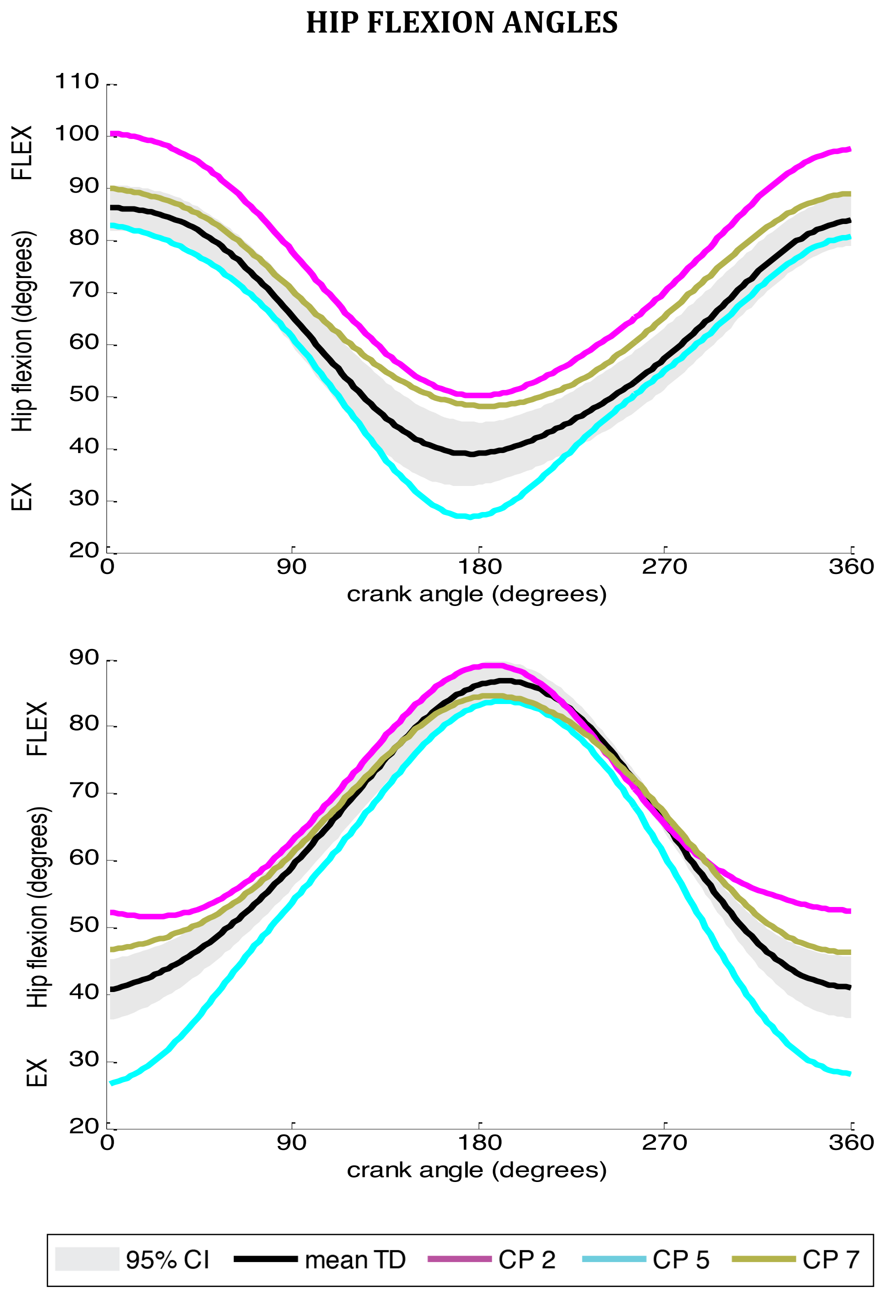


Figure A36: Hip flexion angles at 70 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION ANGLES

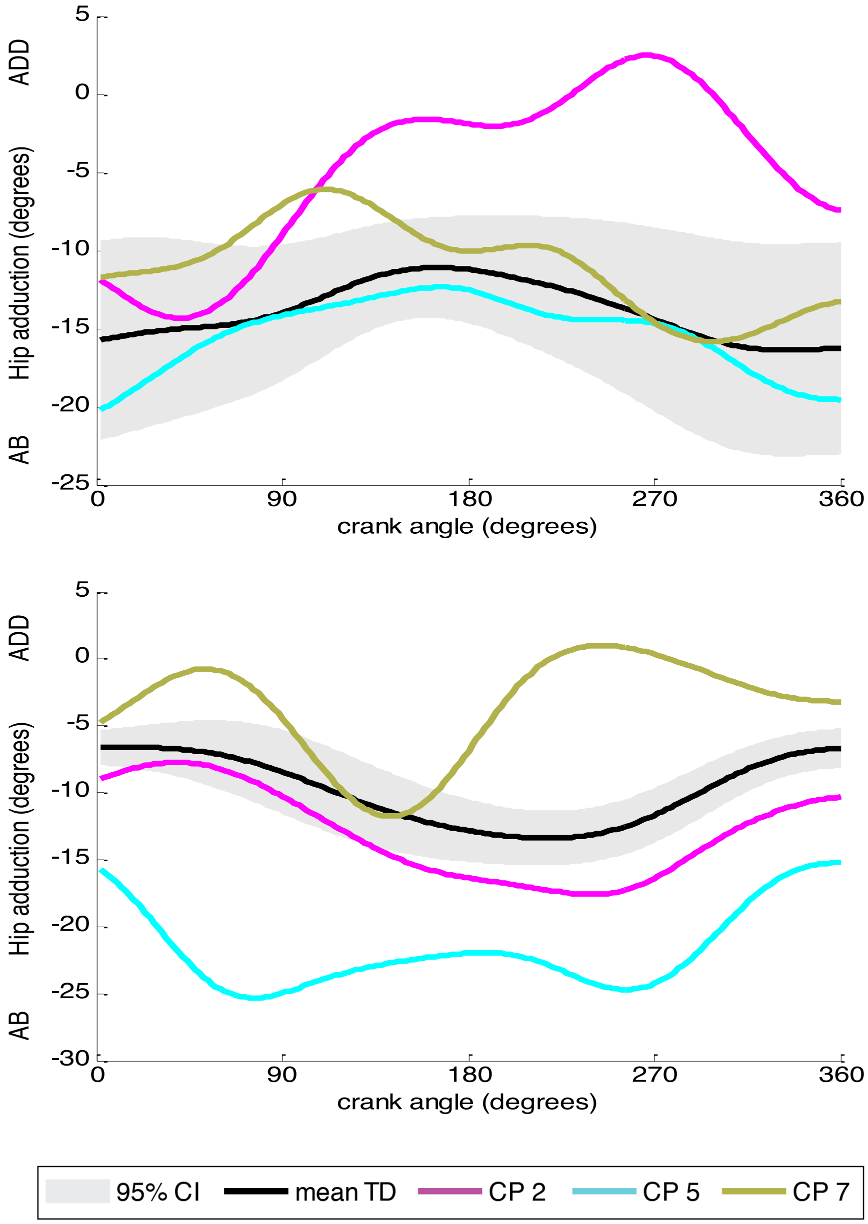


Figure A37: Hip adduction angles at 70 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION ANGLES

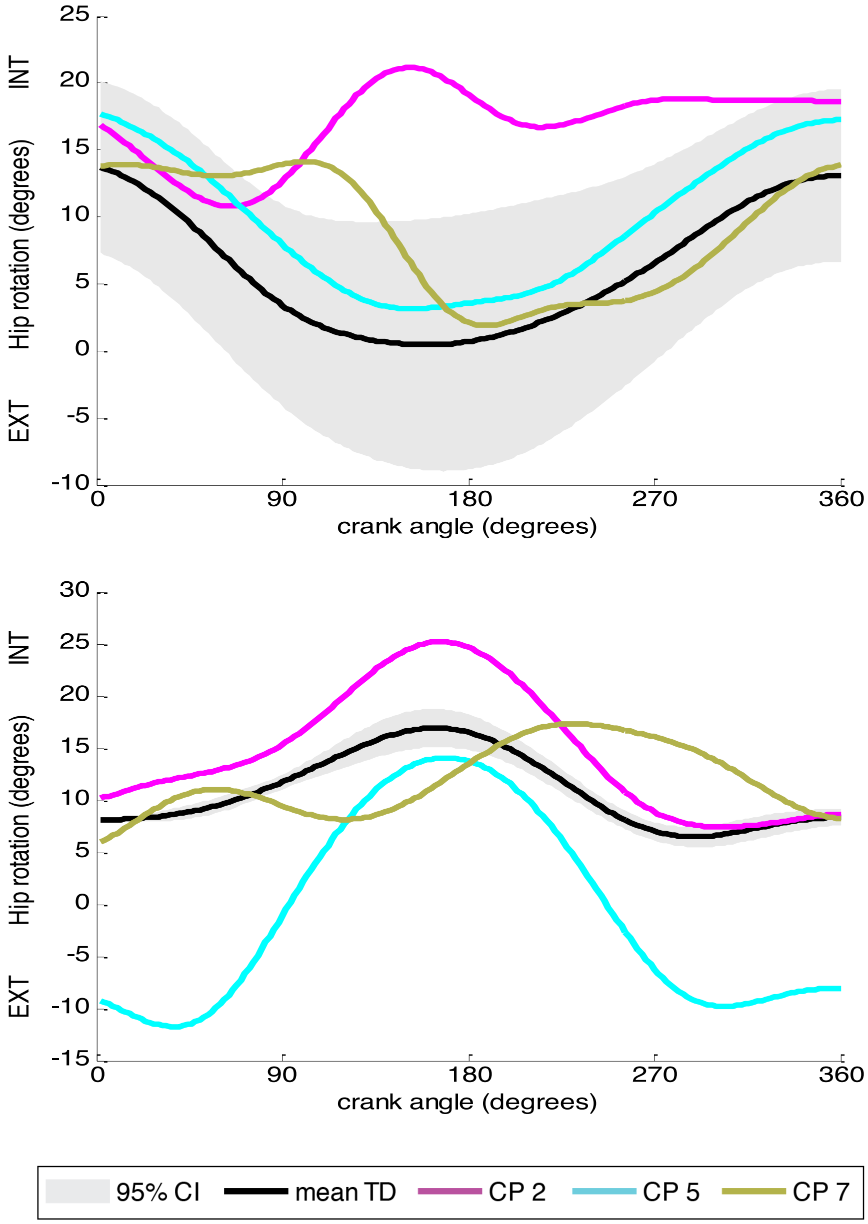


Figure A38: Hip rotation angles at 70 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION ANGLES

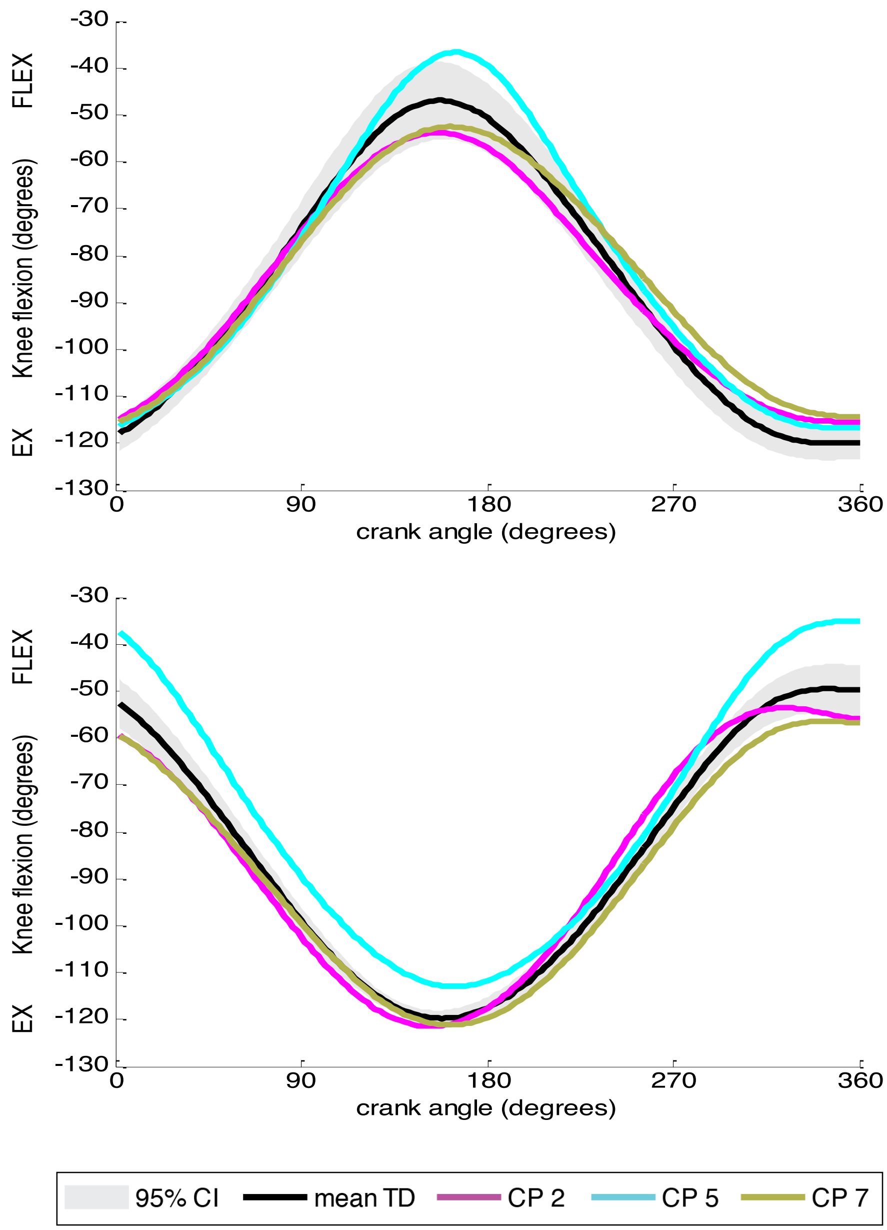


Figure A39: Knee flexion angles at 70 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION ANGLES

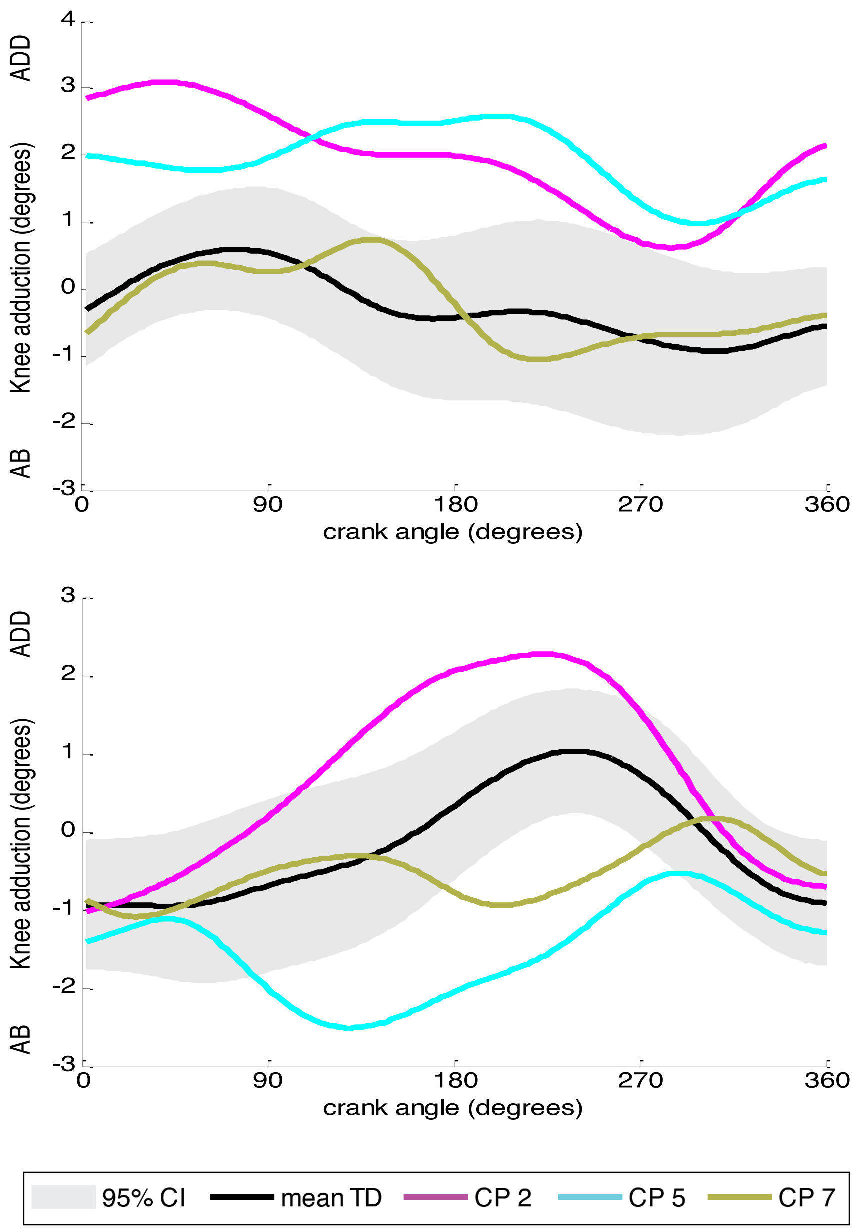


Figure A40: Knee adduction angles at 70 RPM produced by right (top) and left (bottom) leg.

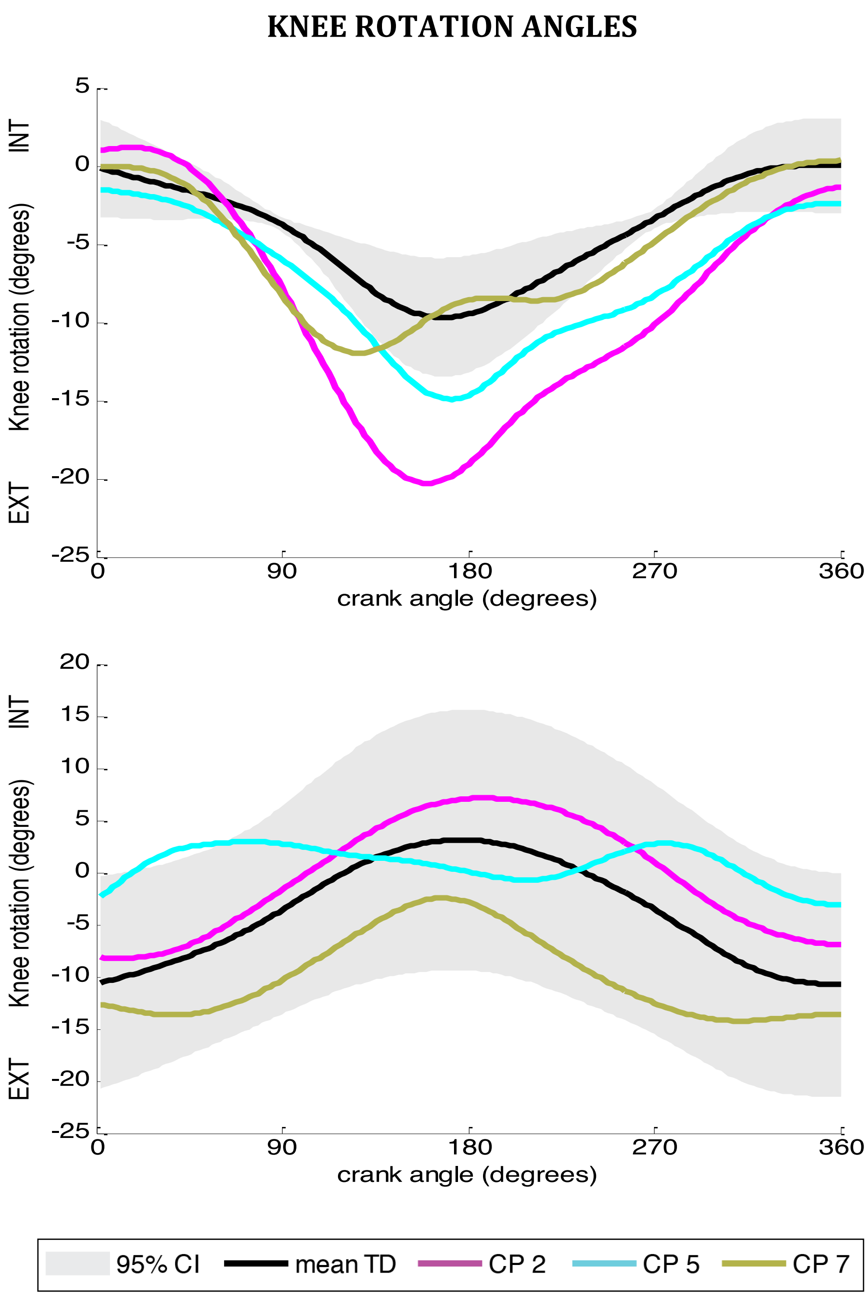


Figure A41: Knee rotation angles at 70 RPM produced by right (top) and left (bottom) leg.

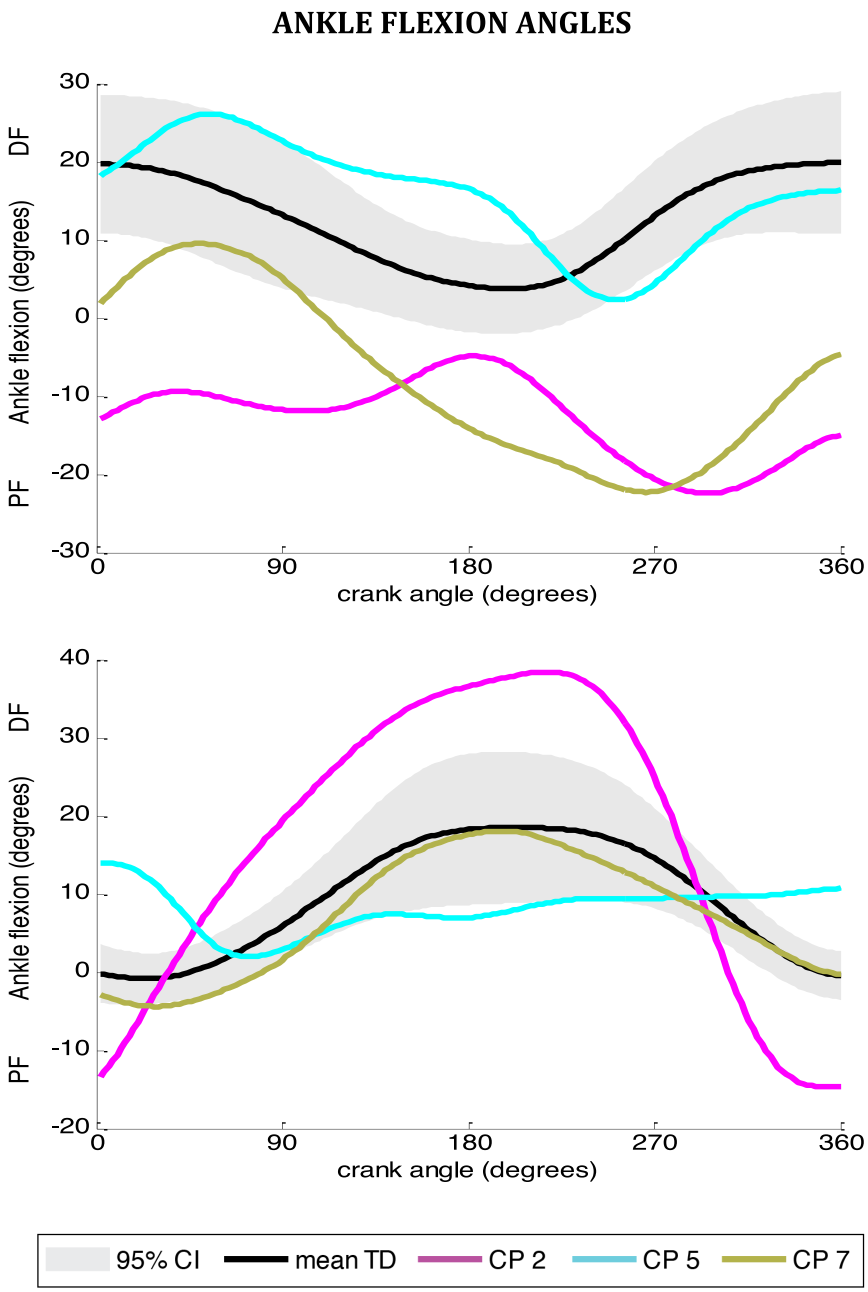


Figure A42: Ankle flexion angles at 70 RPM produced by right (top) and left (bottom) leg.

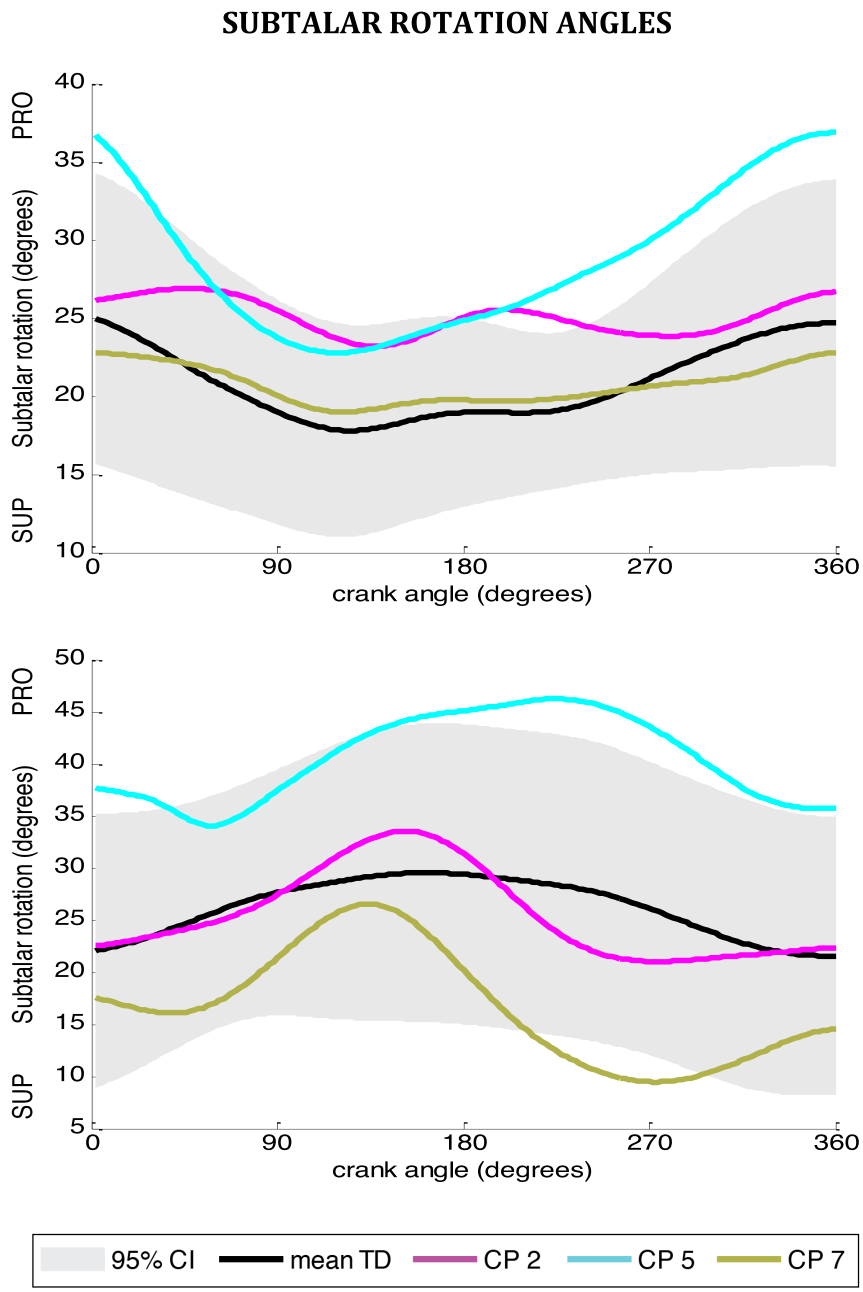


Figure A43: Subtalar rotation angles at 70 RPM produced by right (top) and left (bottom) leg.

APPENDIX Q: JOINT ANGLES AT 80 RPM IN STUDY 2

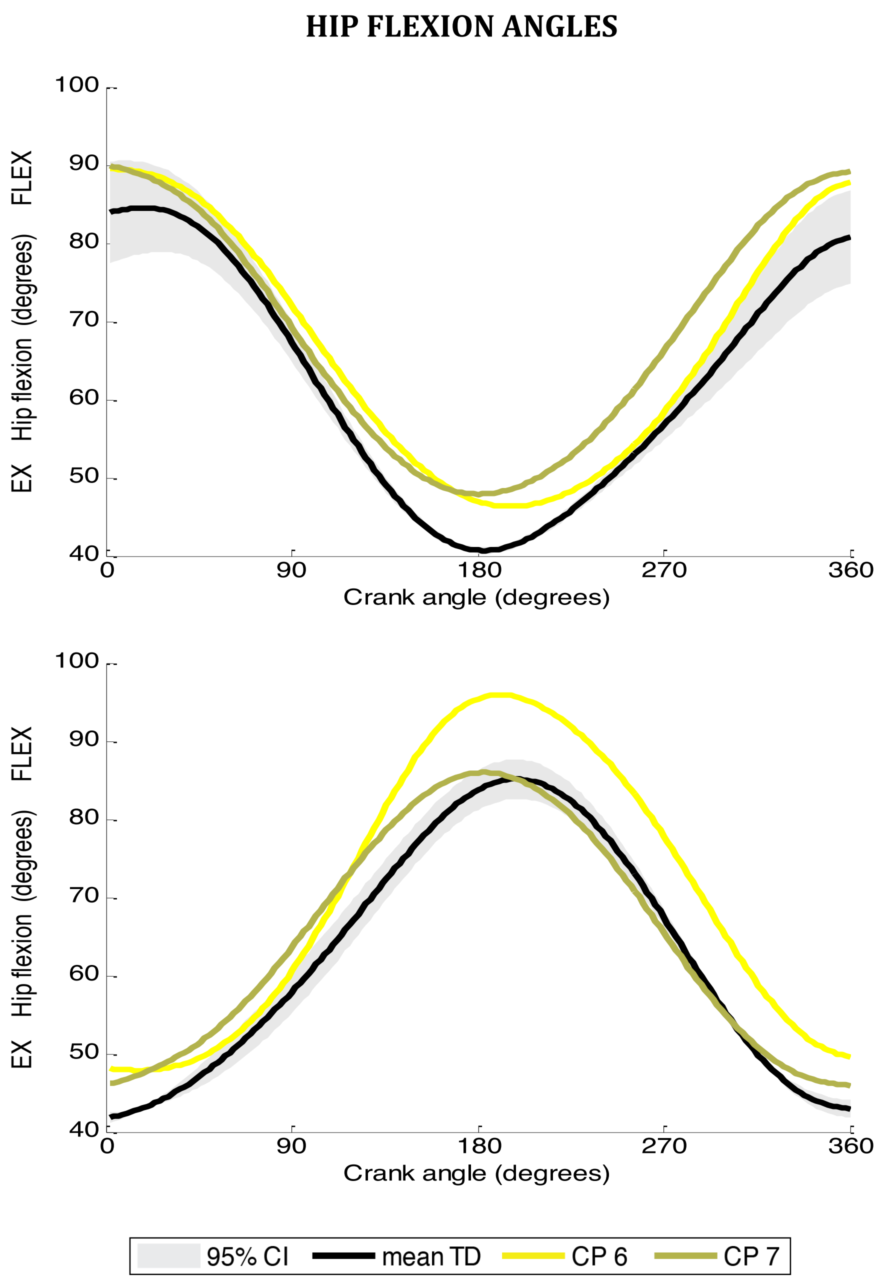


Figure A44: Hip flexion angles at 80 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION ANGLES

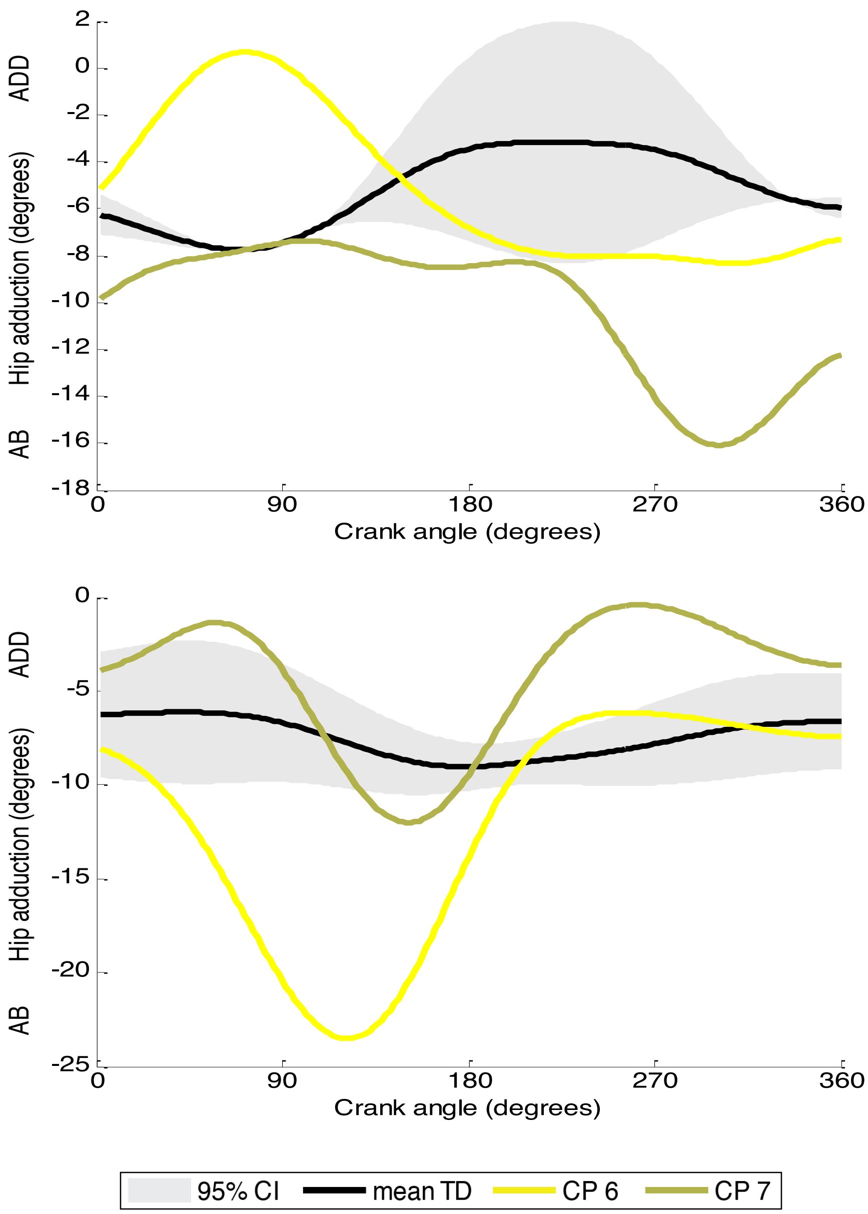


Figure A45: Hip adduction angles at 80 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION ANGLES

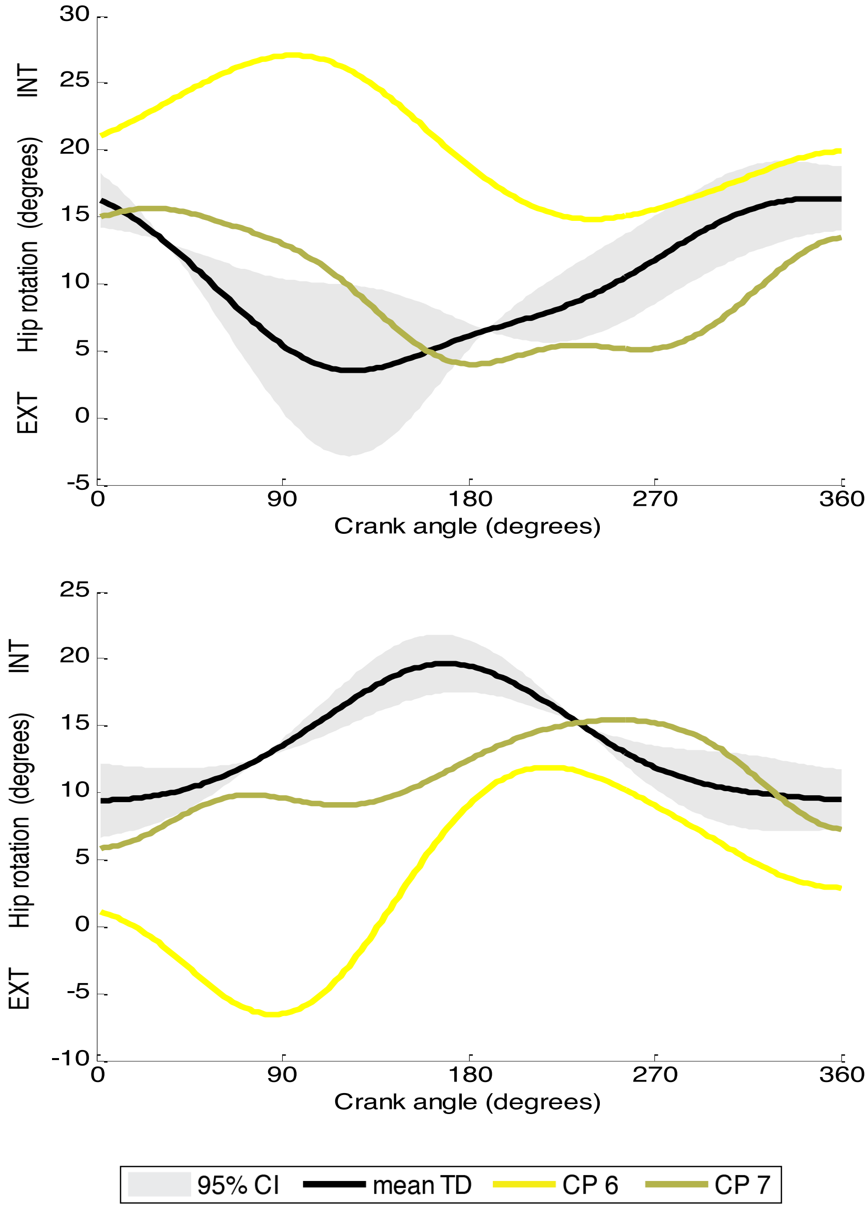


Figure A46: Hip rotation angles at 80 RPM produced by right (top) and left (bottom) leg.

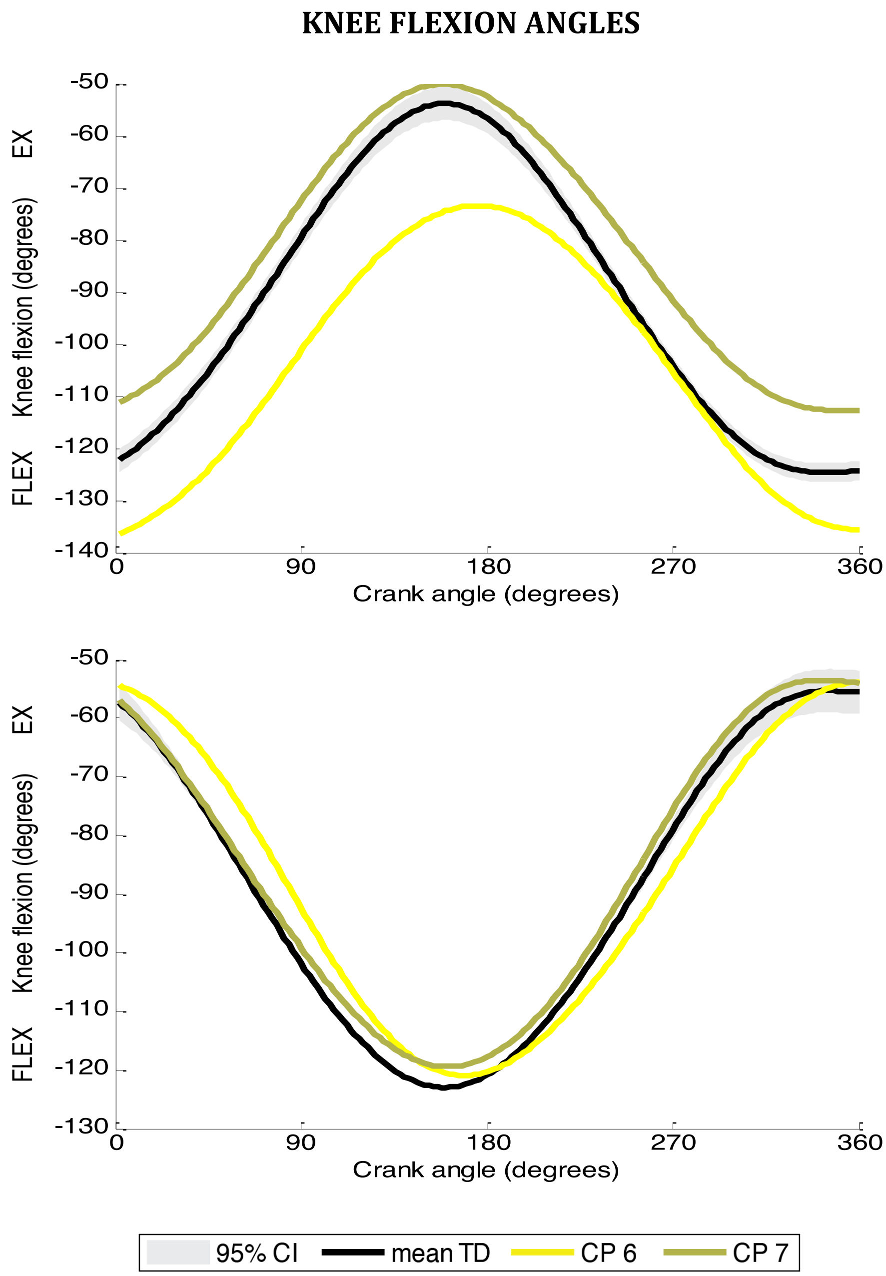


Figure A47: Knee flexion angles at 80 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION ANGLES

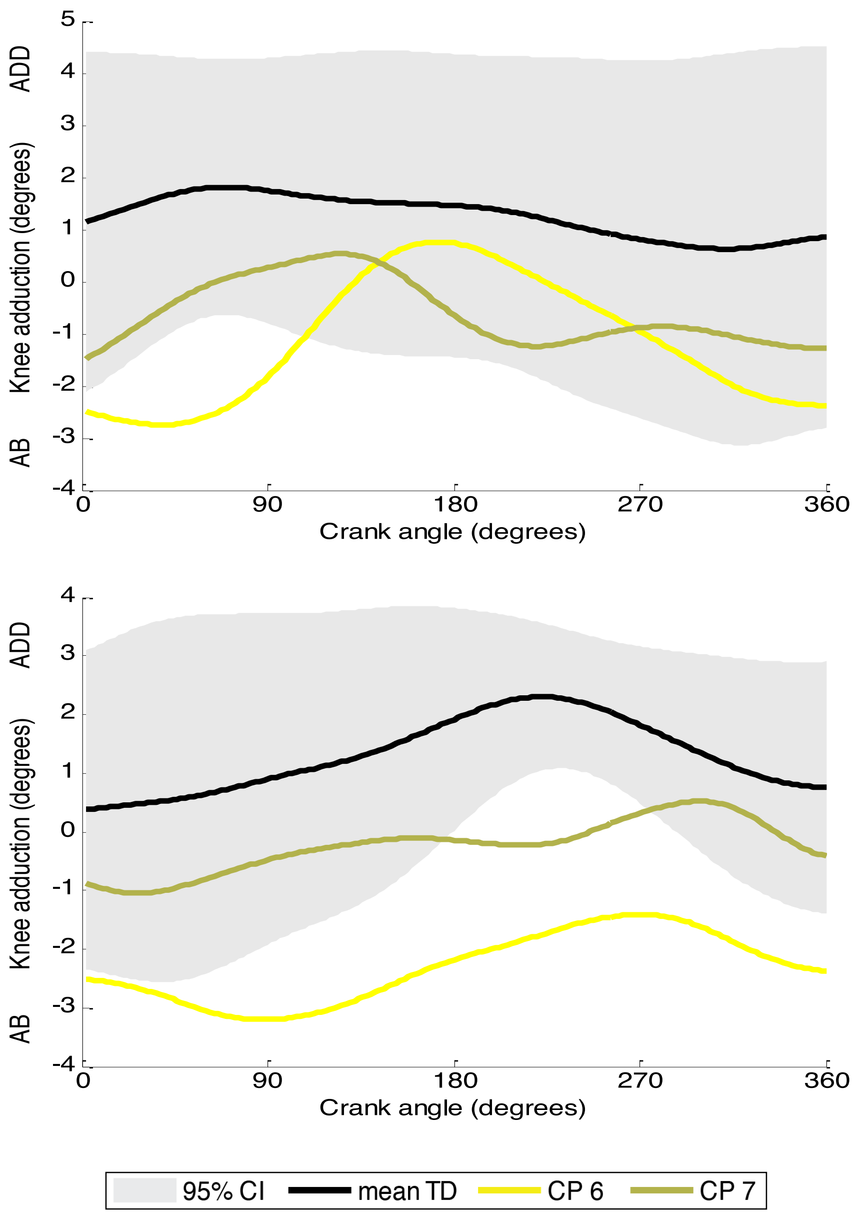


Figure A48: Knee adduction angles at 80 RPM produced by right (top) and left (bottom) leg.

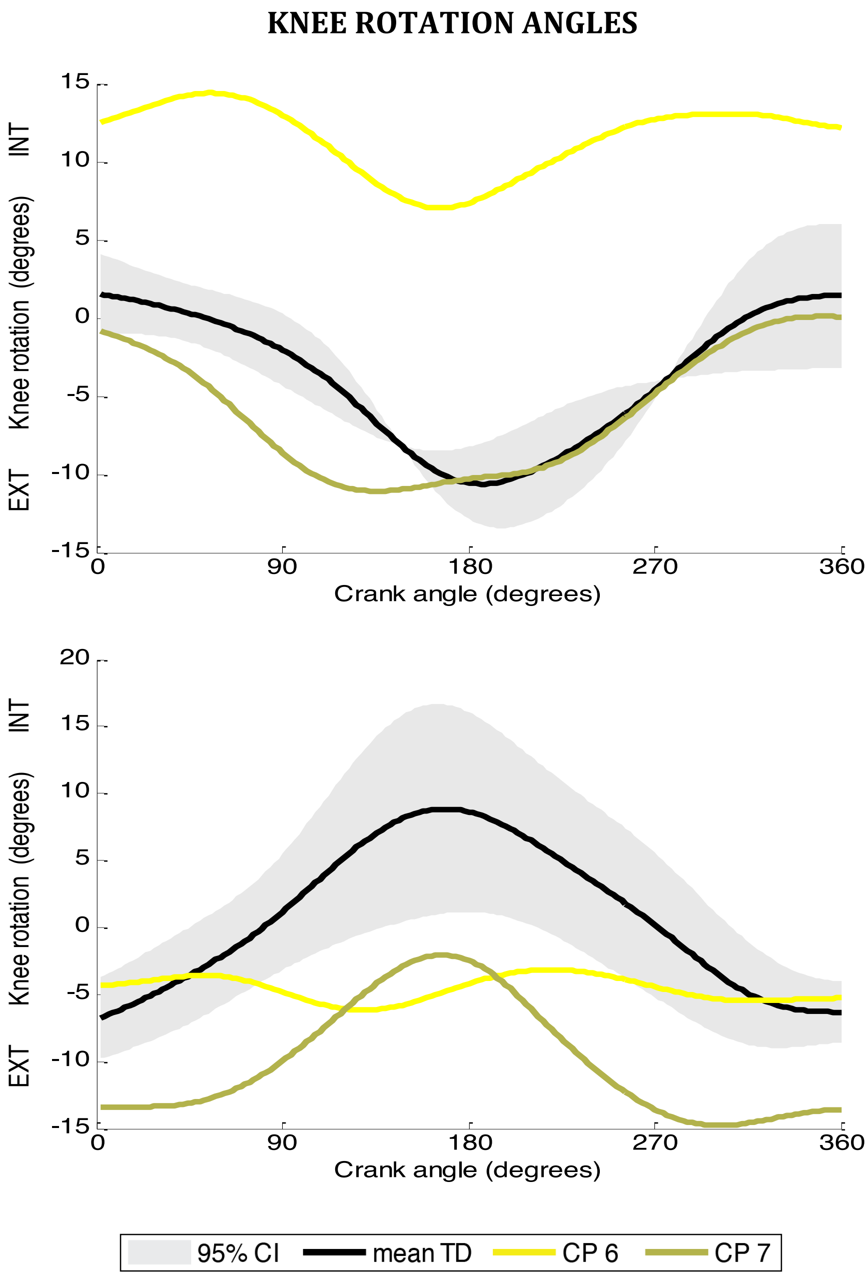


Figure A49: Knee rotation angles at 80 RPM produced by right (top) and left (bottom) leg.

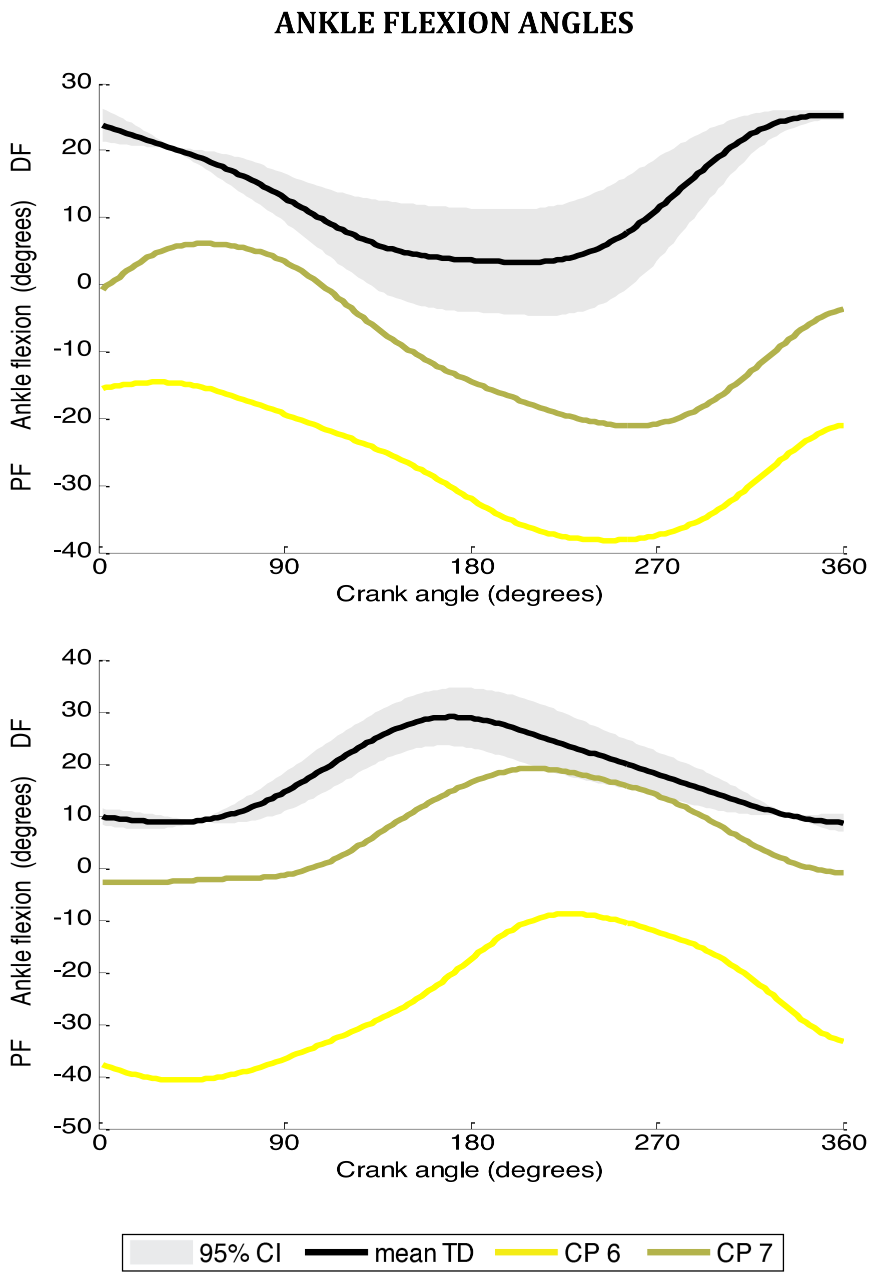


Figure A50: Ankle flexion angles at 80 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION ANGLES

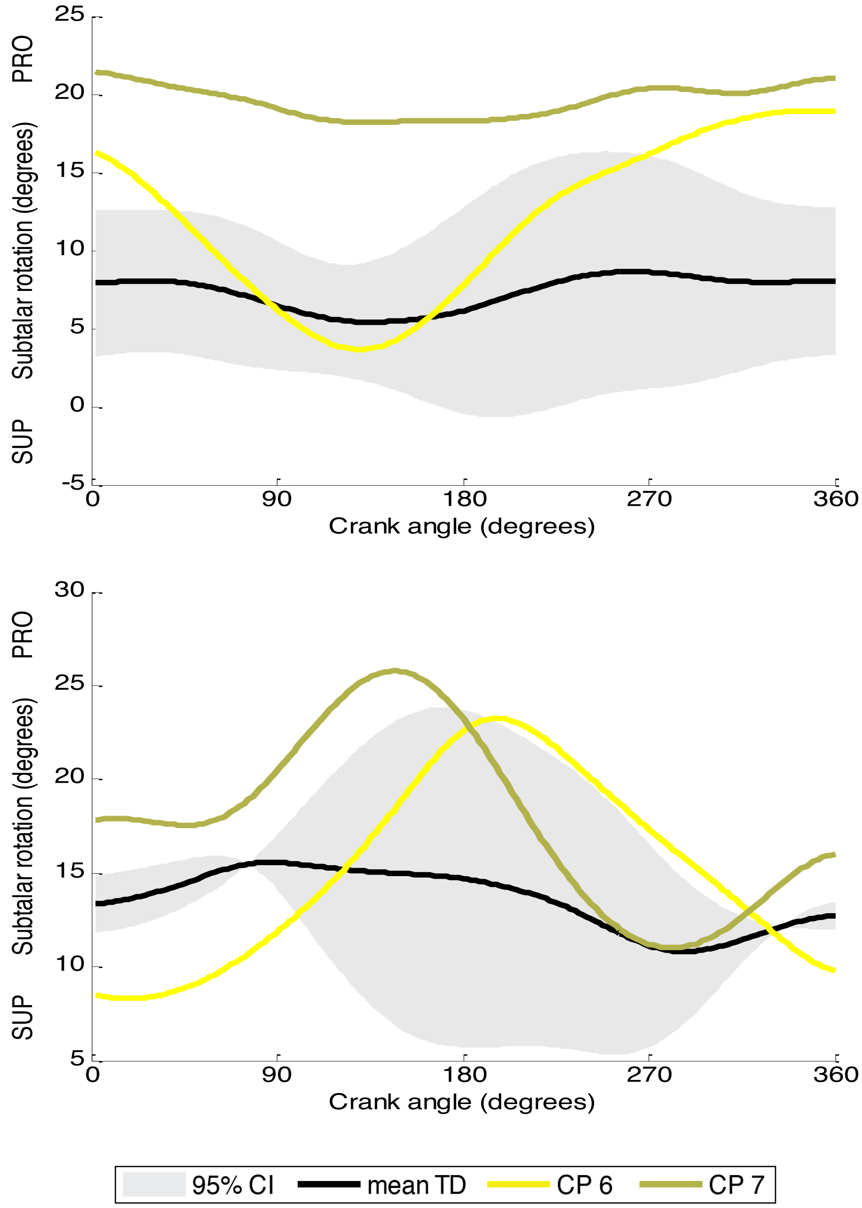


Figure A51: Subtalar rotation angles at 80 RPM produced by right (top) and left (bottom) leg.

APPENDIX R: JERK COST PER PARTICIPANT IN STUDY 2

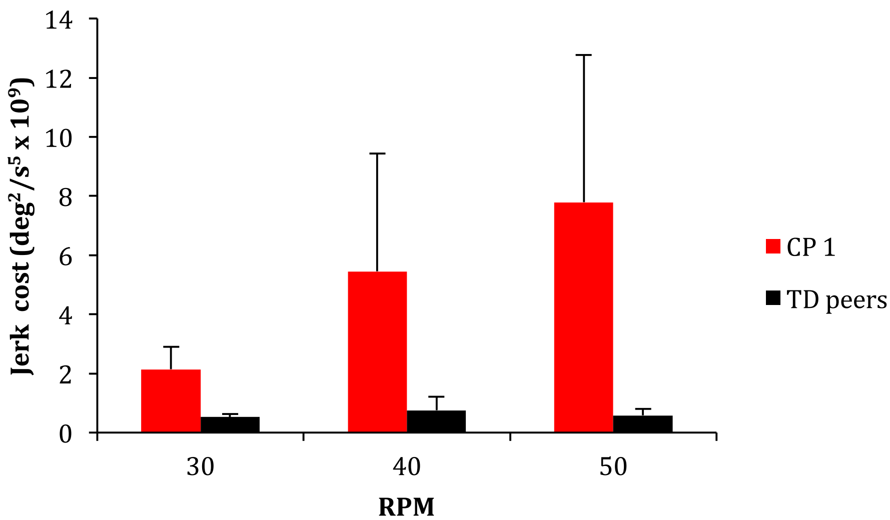


Figure A52: Jerk cost produced by CP 1.

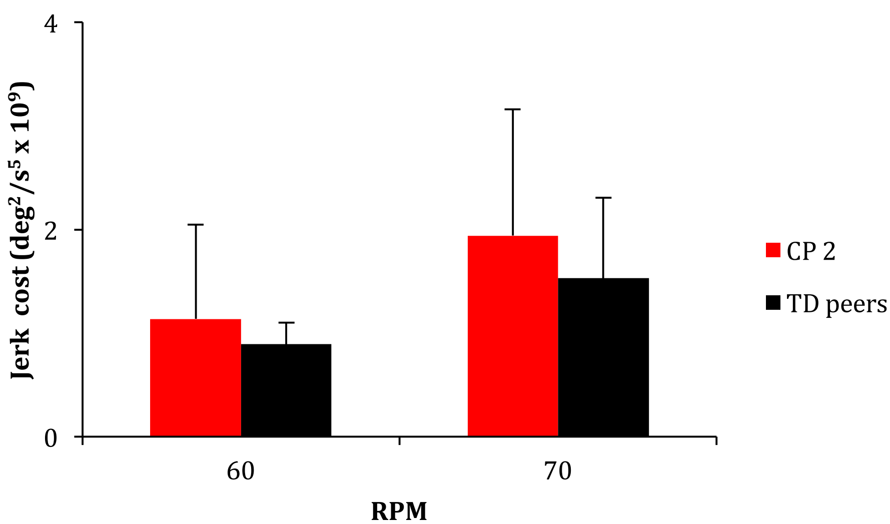


Figure A53: Jerk cost produced by CP 2.

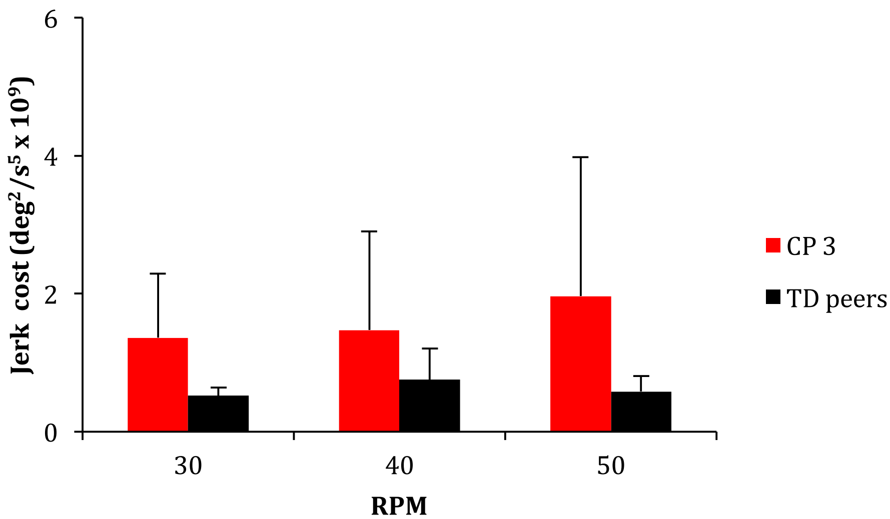


Figure A54: Jerk cost produced by CP 3.

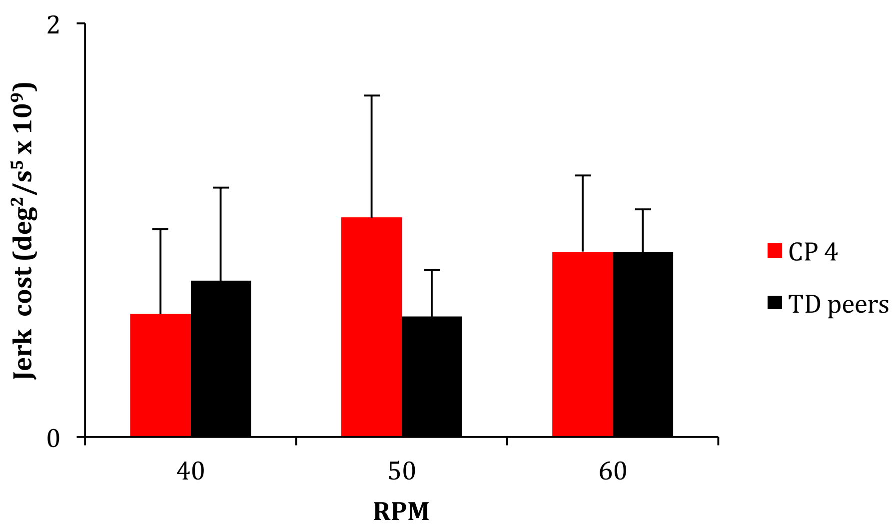


Figure A55: Jerk cost produced by CP 4.

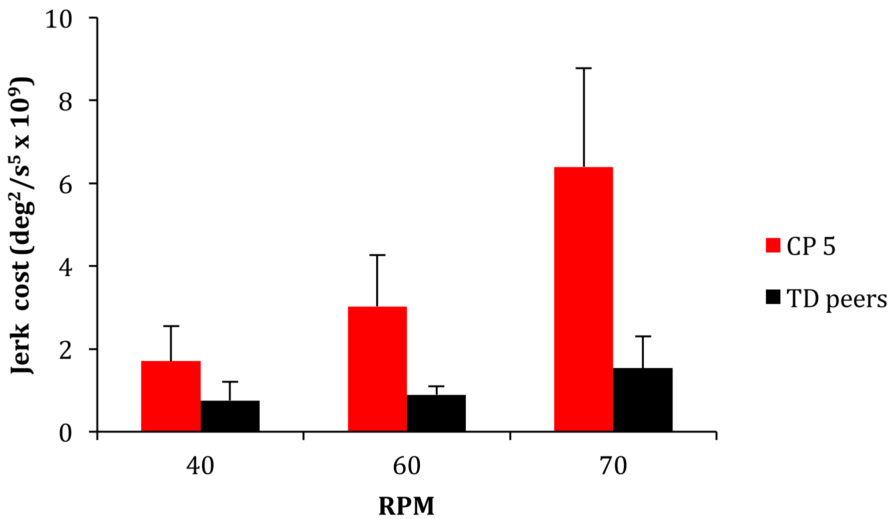


Figure A56: Jerk cost produced by CP 5.

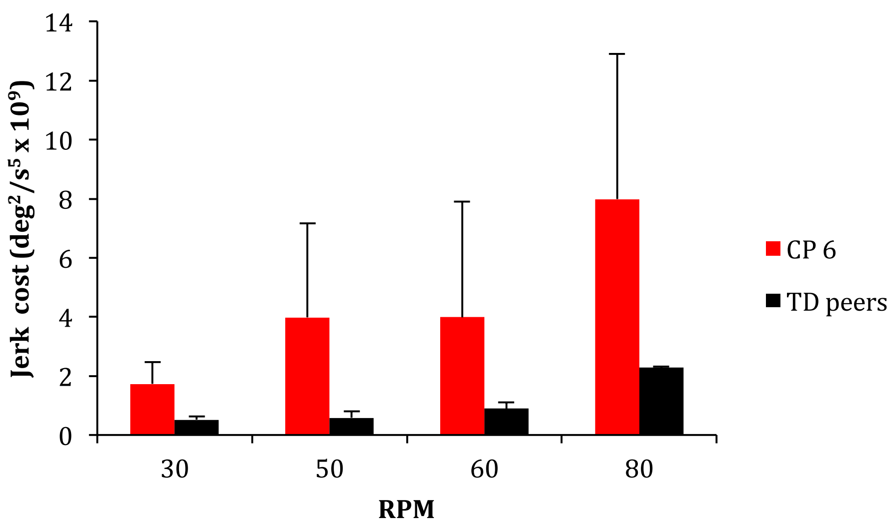


Figure A57: Jerk cost produced by CP 6.

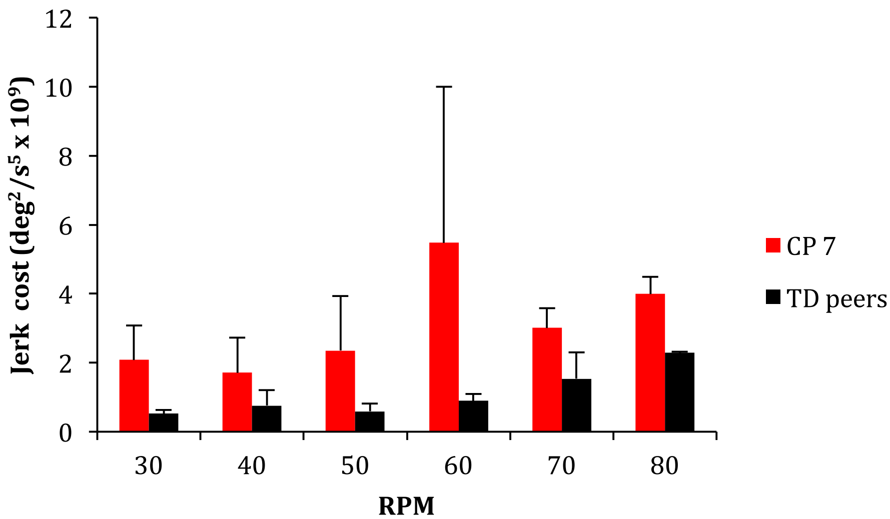


Figure A58: Jerk cost produced by CP 7.

APPENDIX S: PEDAL FORCES AT 30 RPM IN STUDY 2

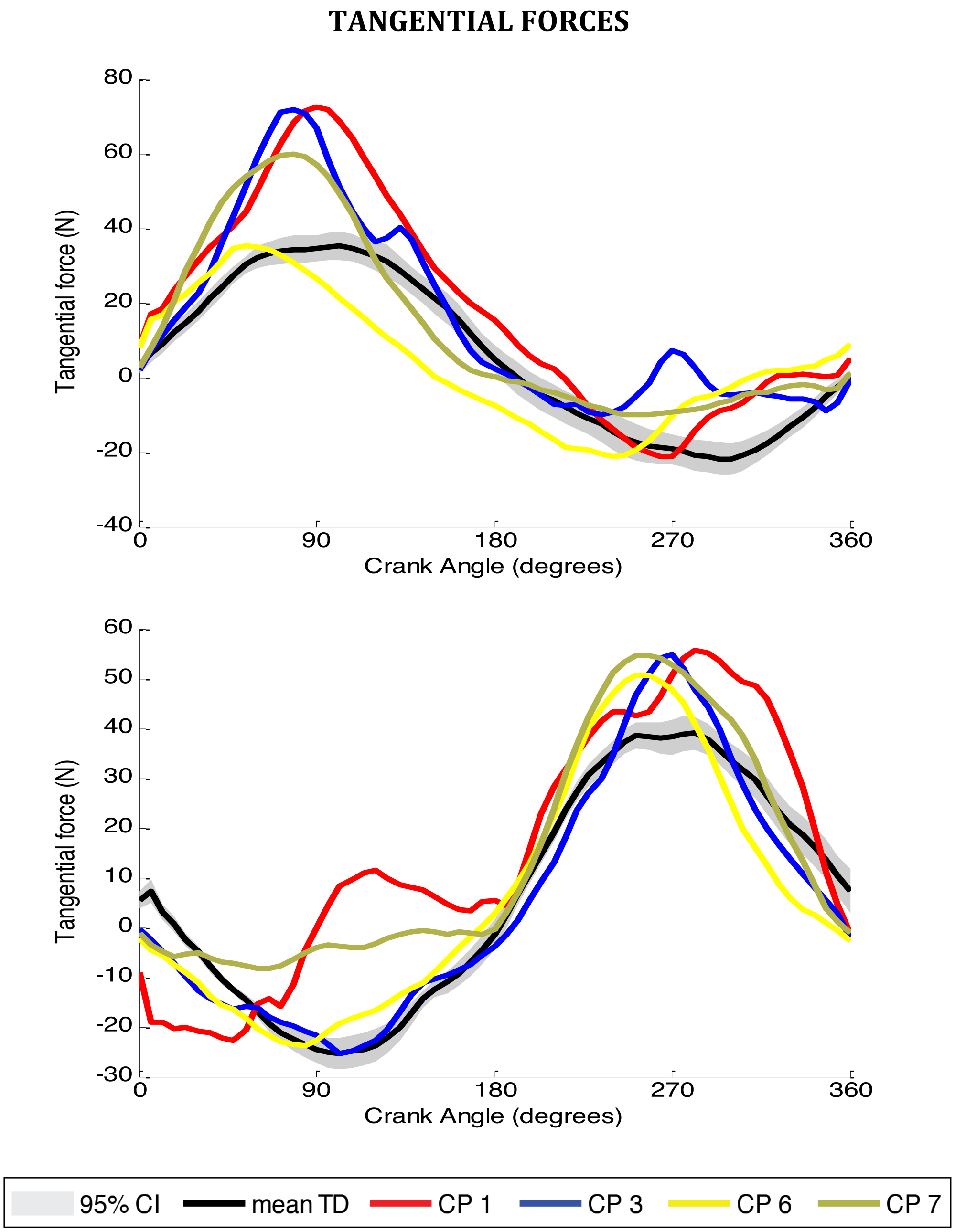


Figure A59: Tangential forces at 30 RPM produced by right (top) and left (bottom) leg.

NORMAL FORCES

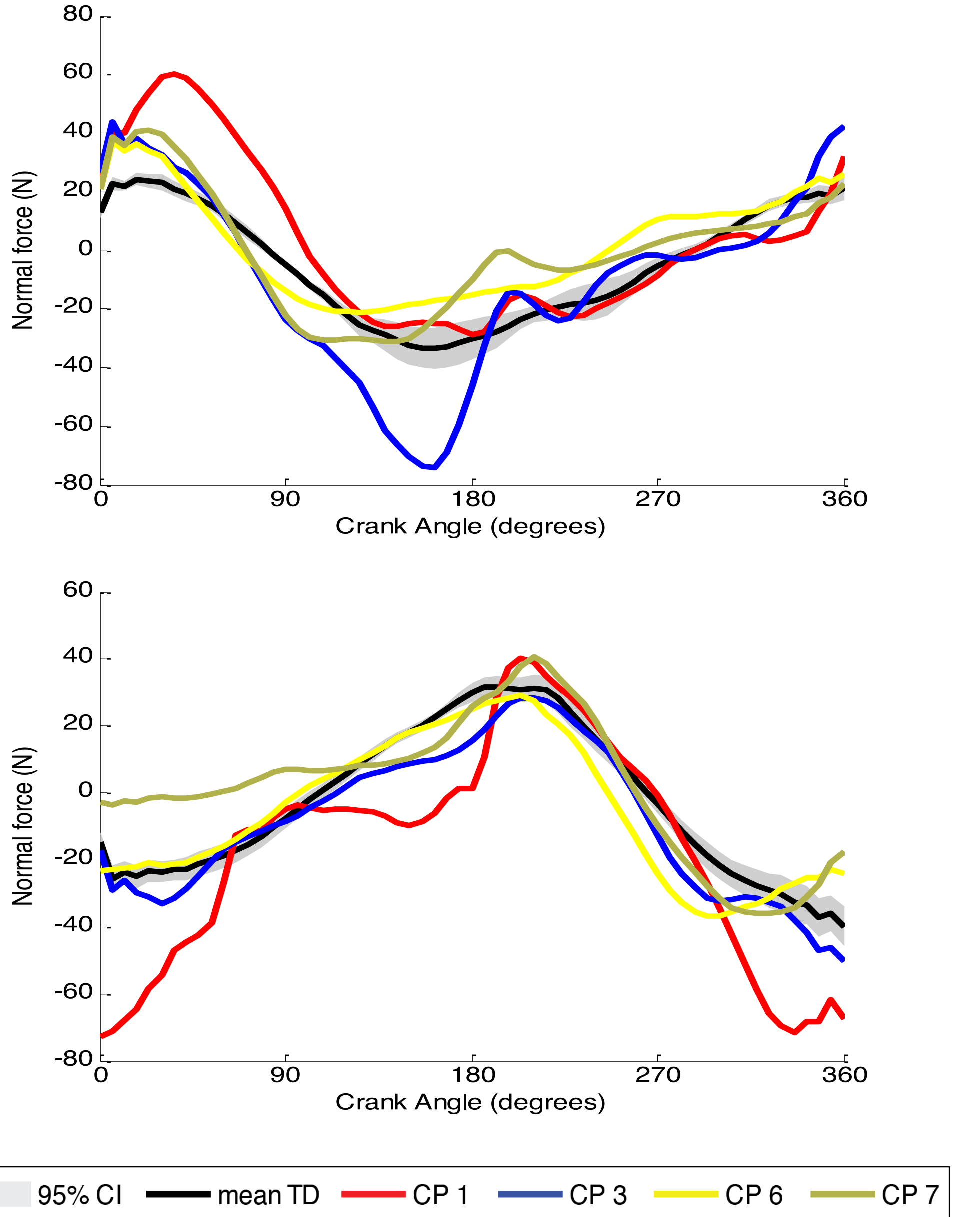


Figure A60: Normal forces at 30 RPM produced by right (top) and left (bottom) leg.

SHEAR FORCES

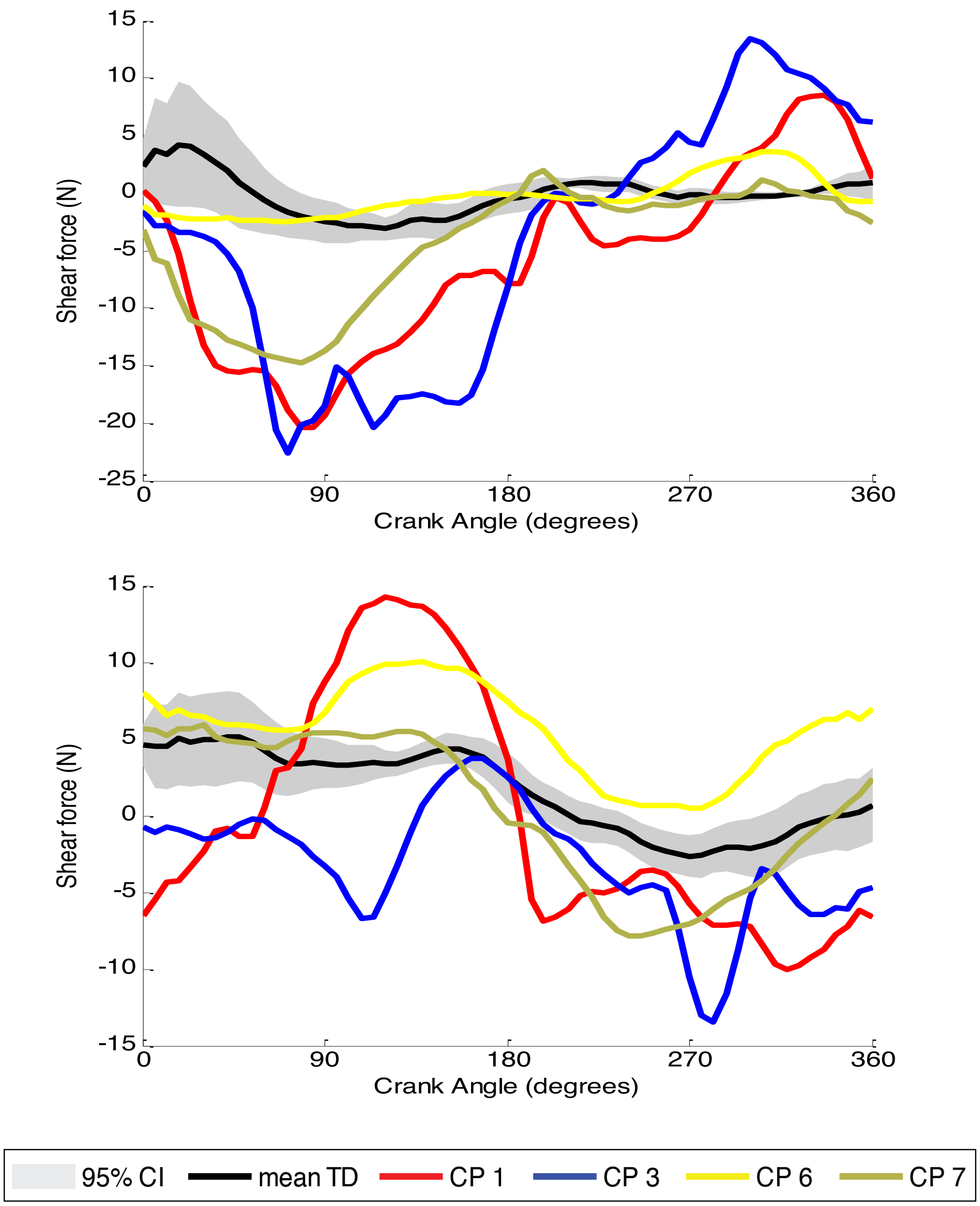


Figure A61: Shear forces at 30 RPM produced by right (top) and left (bottom) leg.

APPENDIX T: PEDAL FORCES AT 40 RPM IN STUDY 2

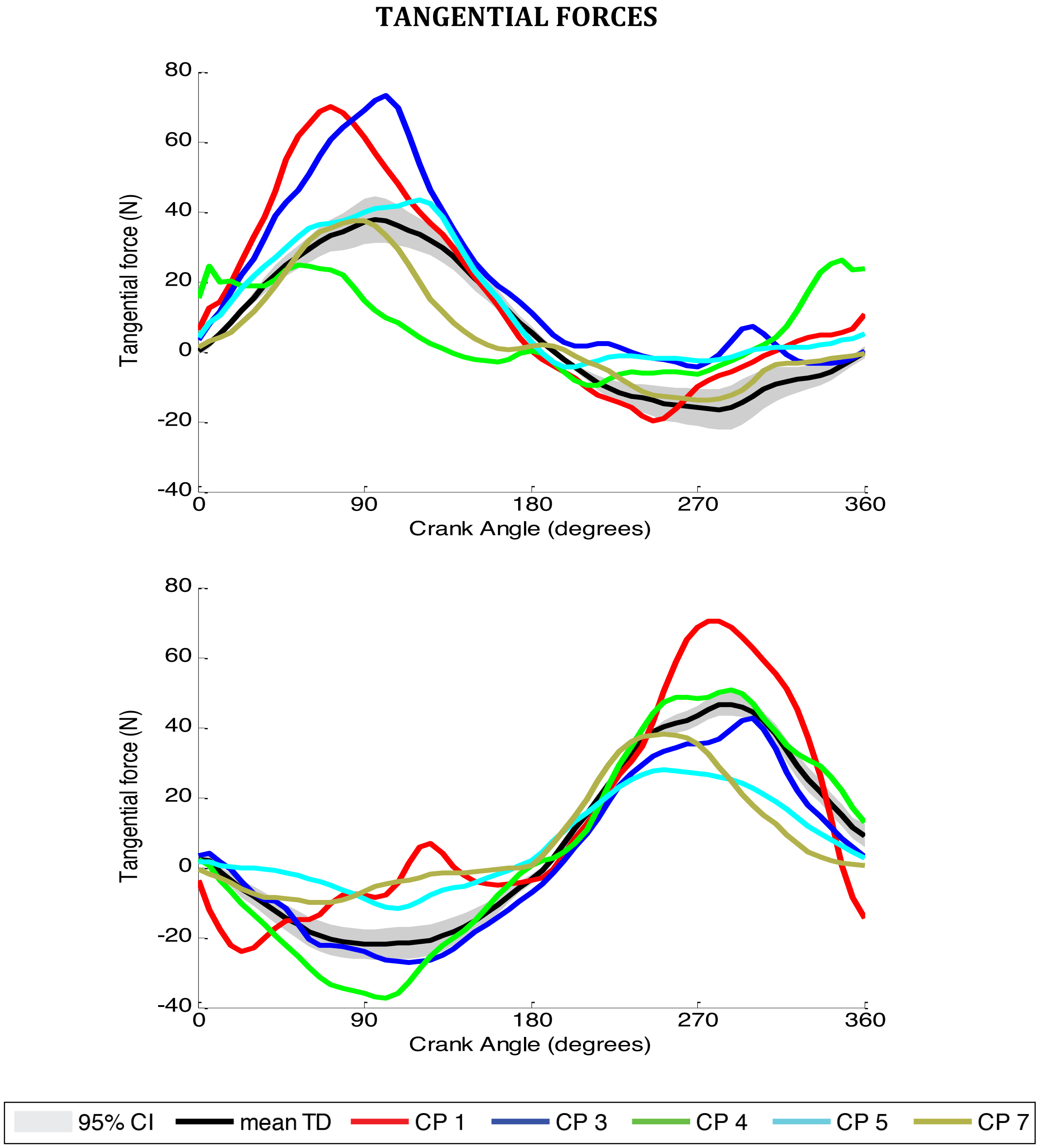


Figure A62: Tangential forces at 40 RPM produced by right (top) and left (bottom) leg.

NORMAL FORCES

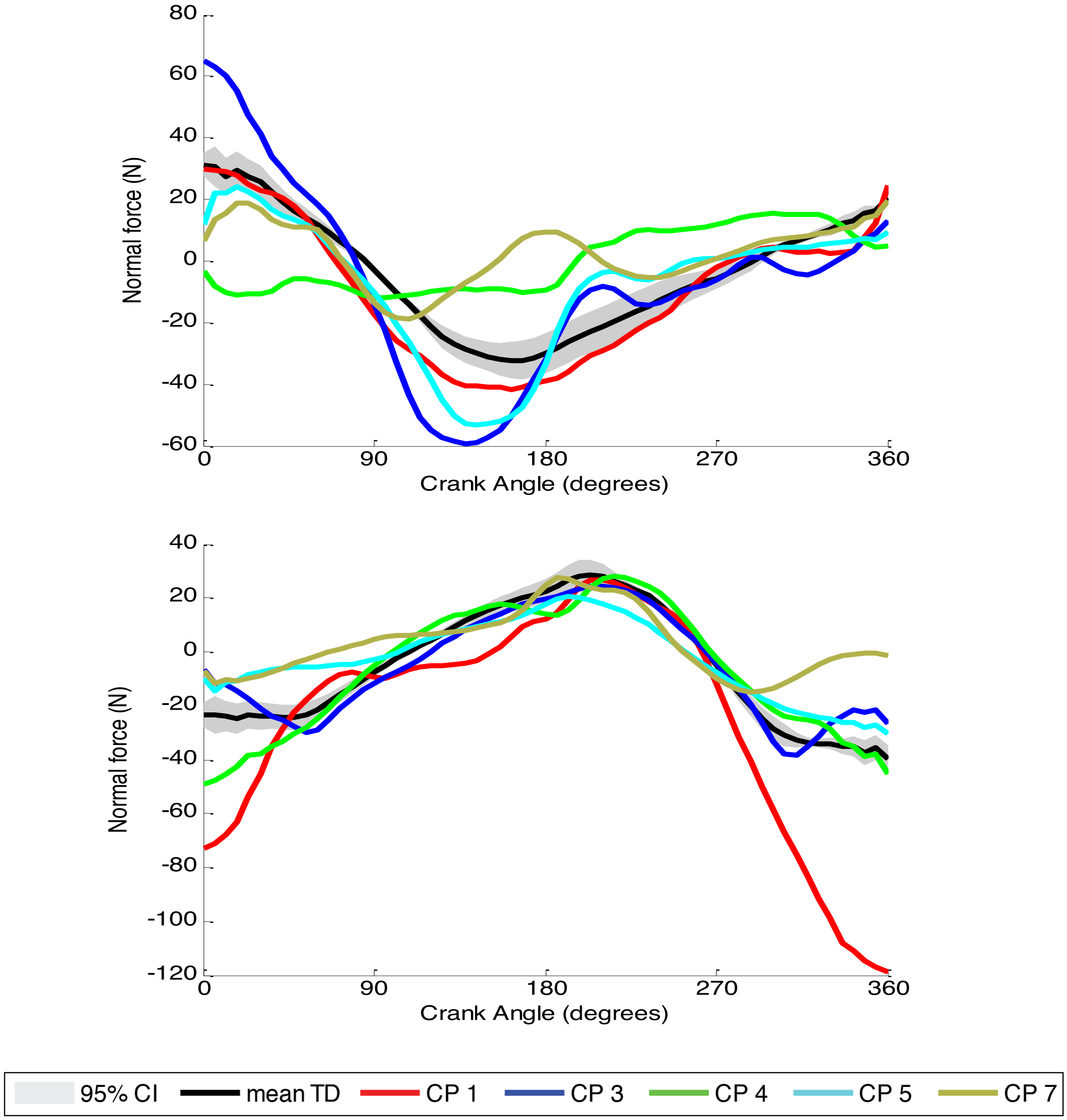


Figure A63: Normal forces at 40 RPM produced by right (top) and left (bottom) leg.

SHEAR FORCES

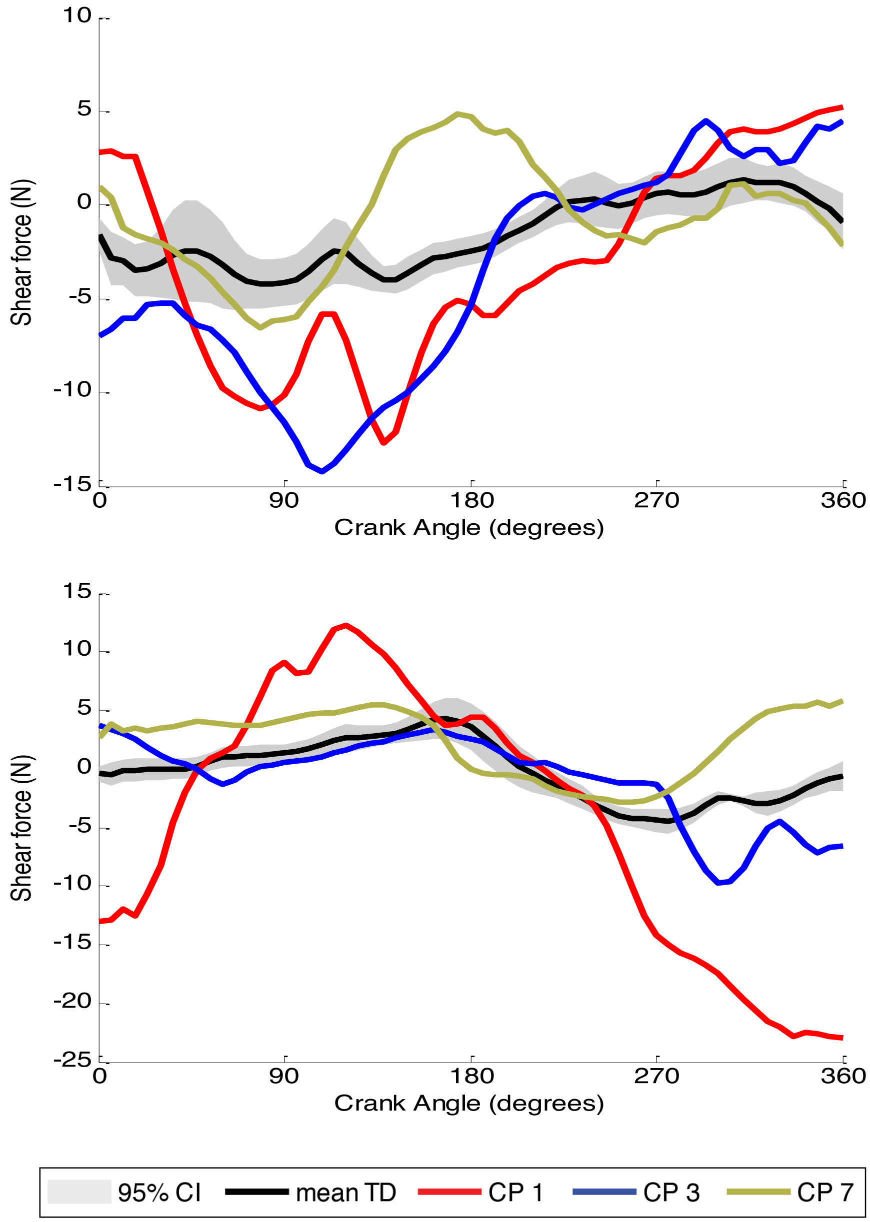


Figure A64: Shear forces at 40 RPM produced by right (top) and left (bottom) leg.

APPENDIX U: PEDAL FORCES AT 50 RPM IN STUDY 2

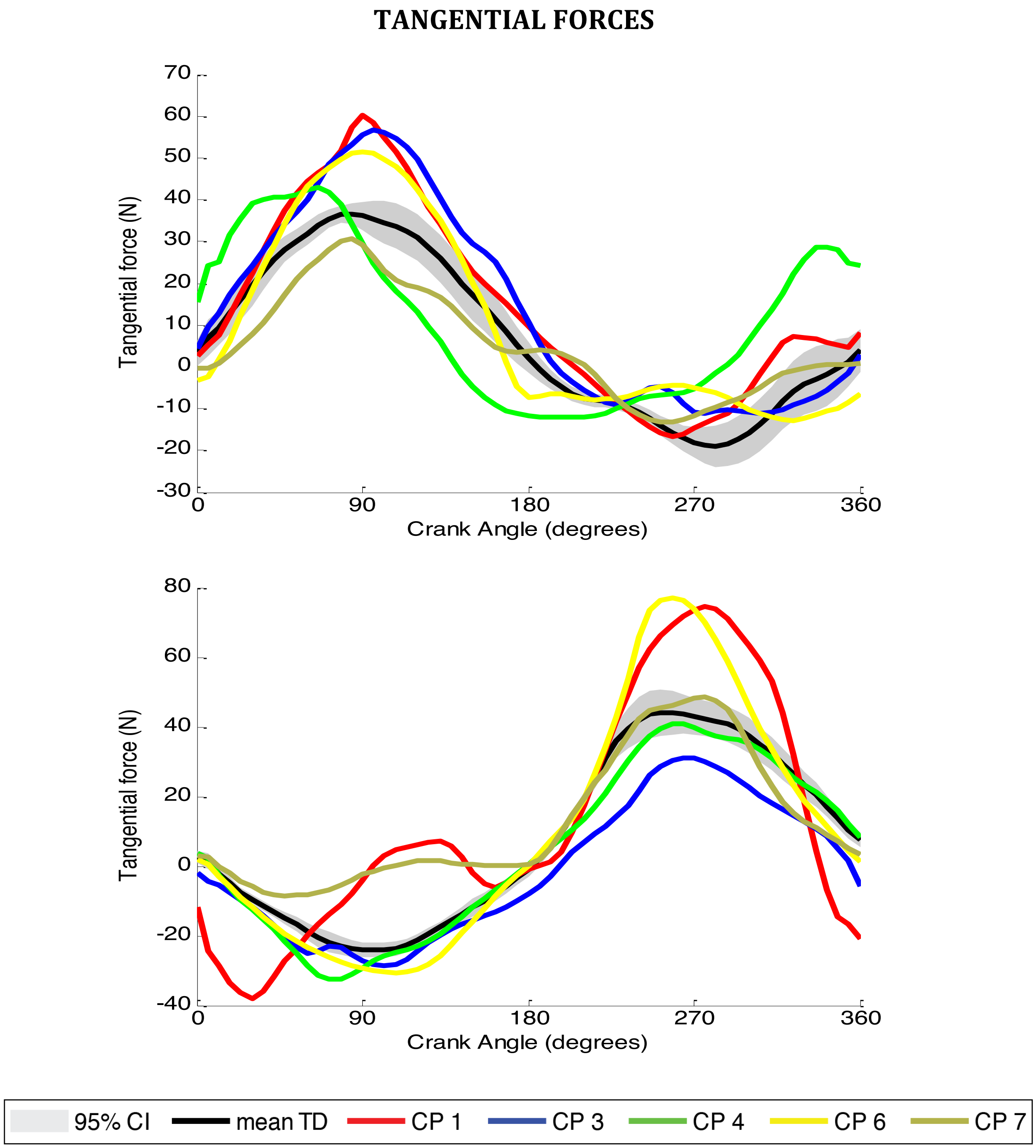


Figure A65: Tangential forces at 50 RPM produced by right (top) and left (bottom) leg.

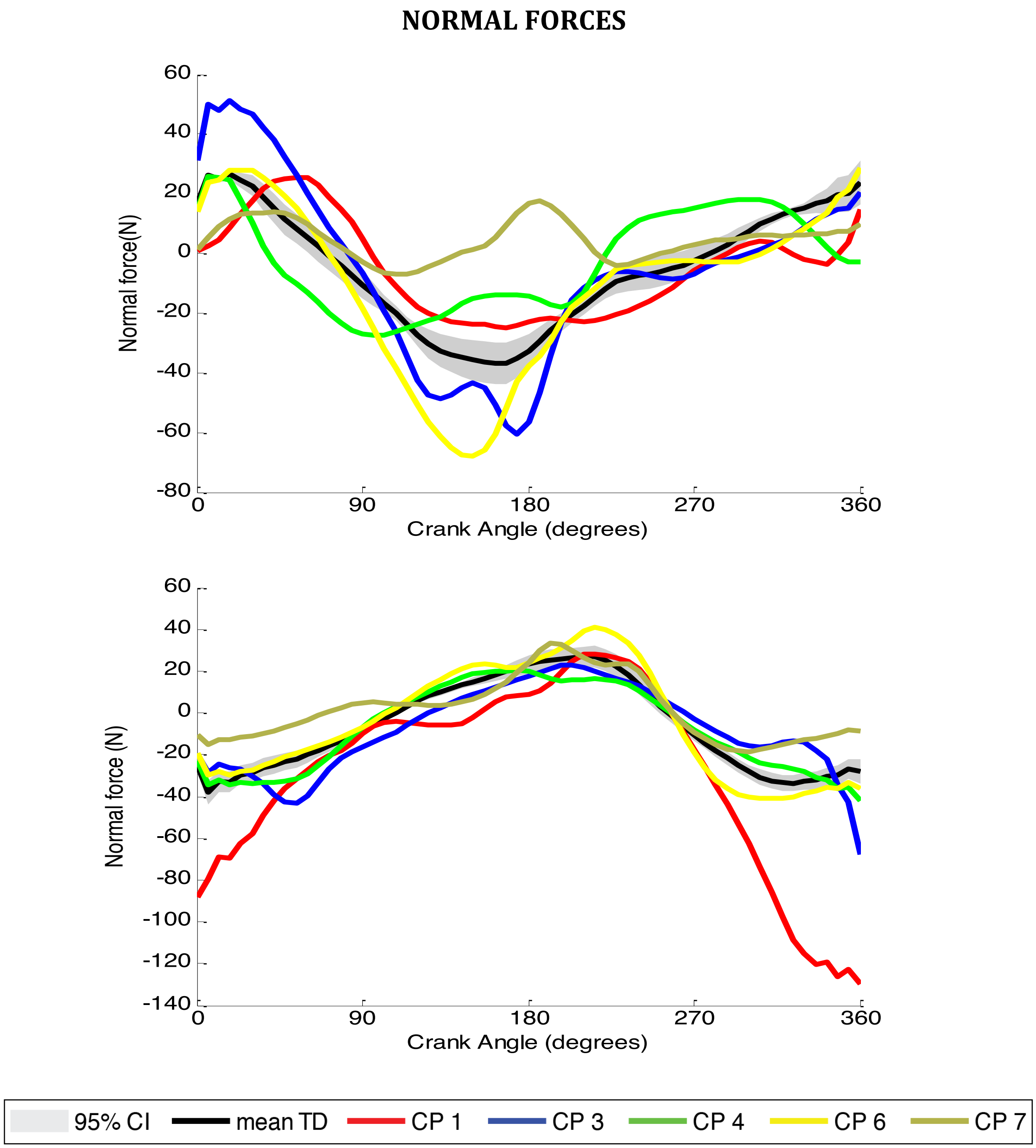


Figure A66: Normal forces at 50 RPM produced by right (top) and left (bottom) leg.

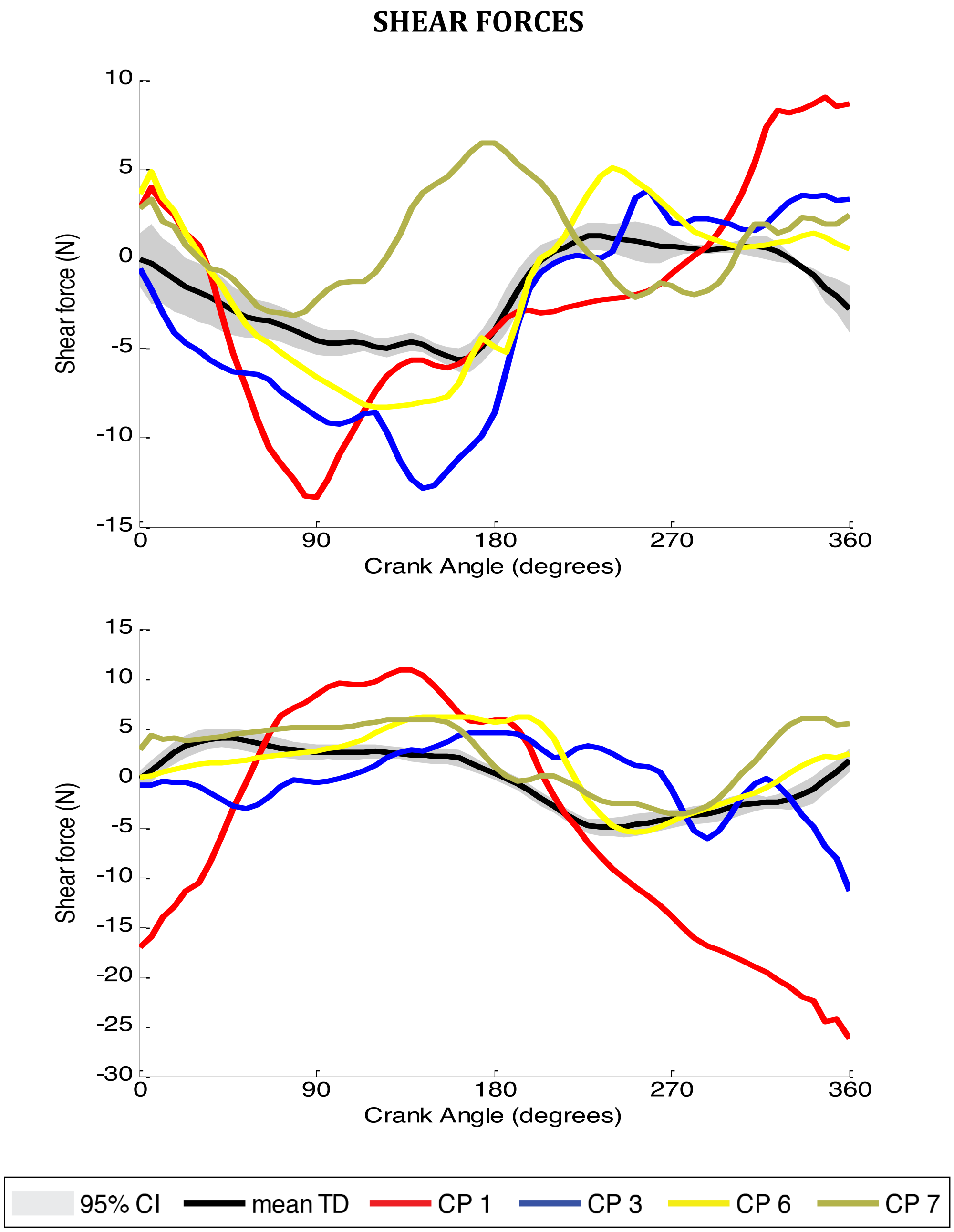


Figure A67: Shear forces at 50 RPM produced by right (top) and left (bottom) leg.

APPENDIX V: PEDAL FORCES AT 60 RPM IN STUDY 2

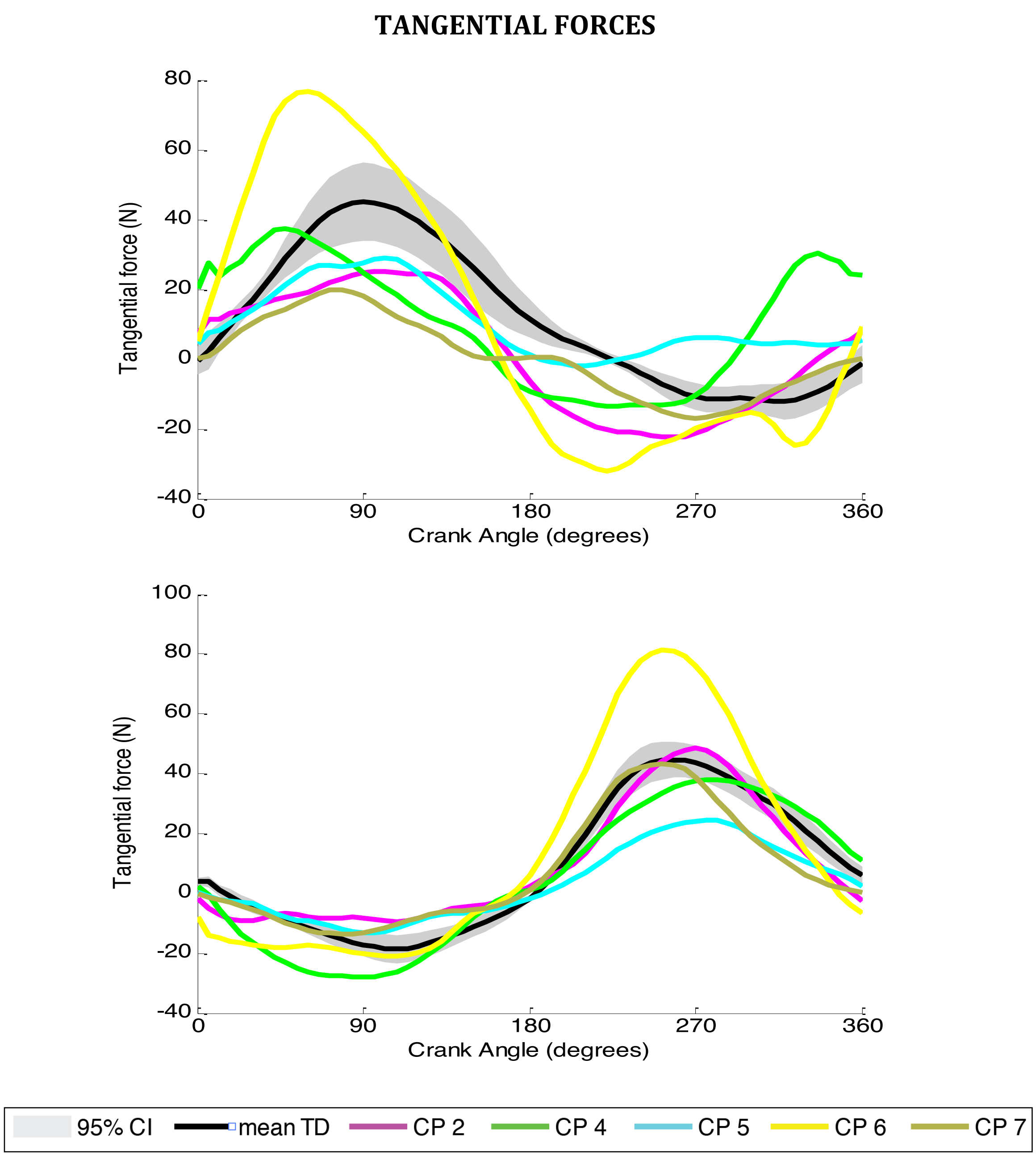


Figure A68: Tangential forces at 60 RPM produced by right (top) and left (bottom) leg.

NORMAL FORCES

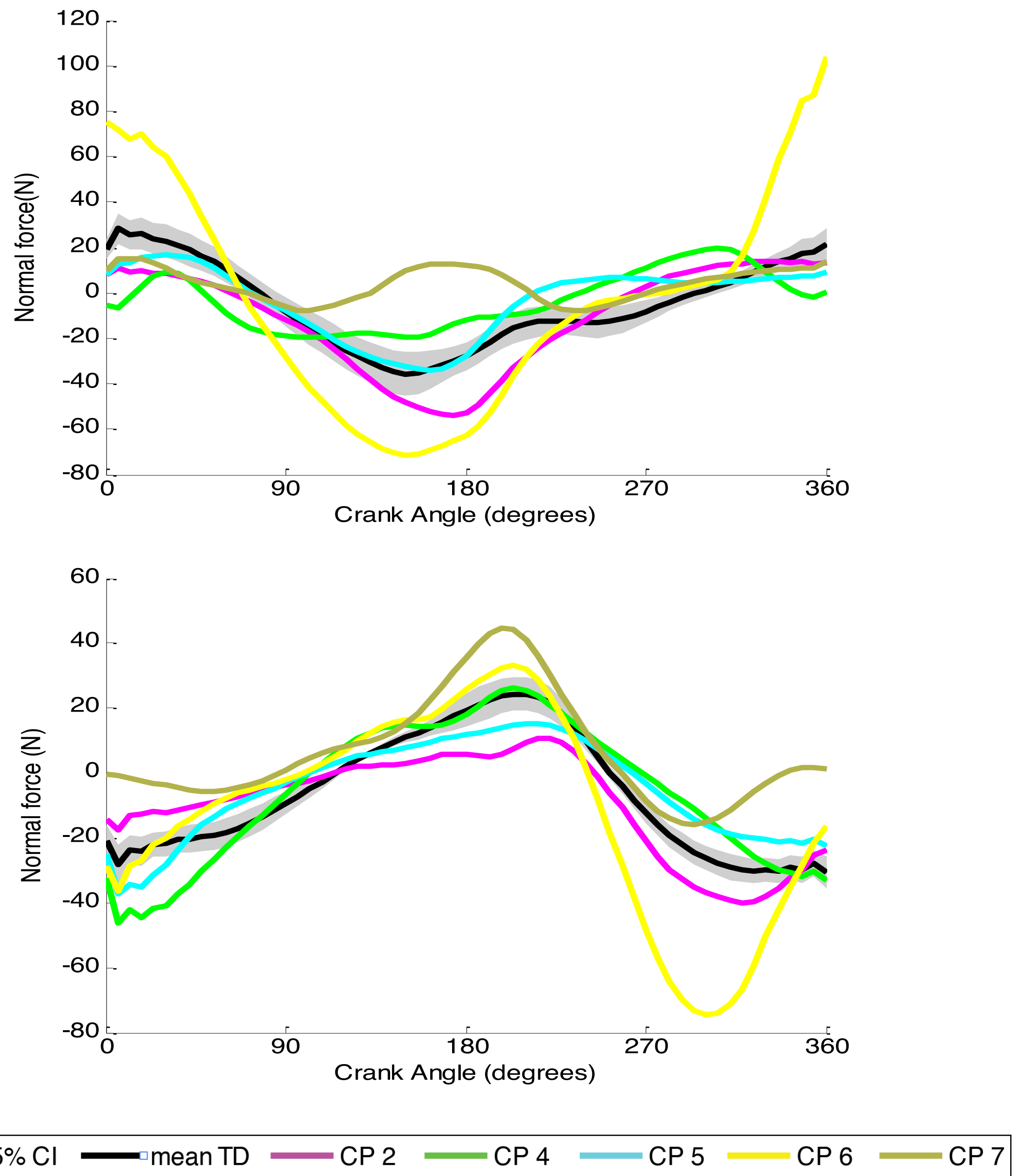


Figure A69: Normal forces at 60 RPM produced by right (top) and left (bottom) leg.

SHEAR FORCES

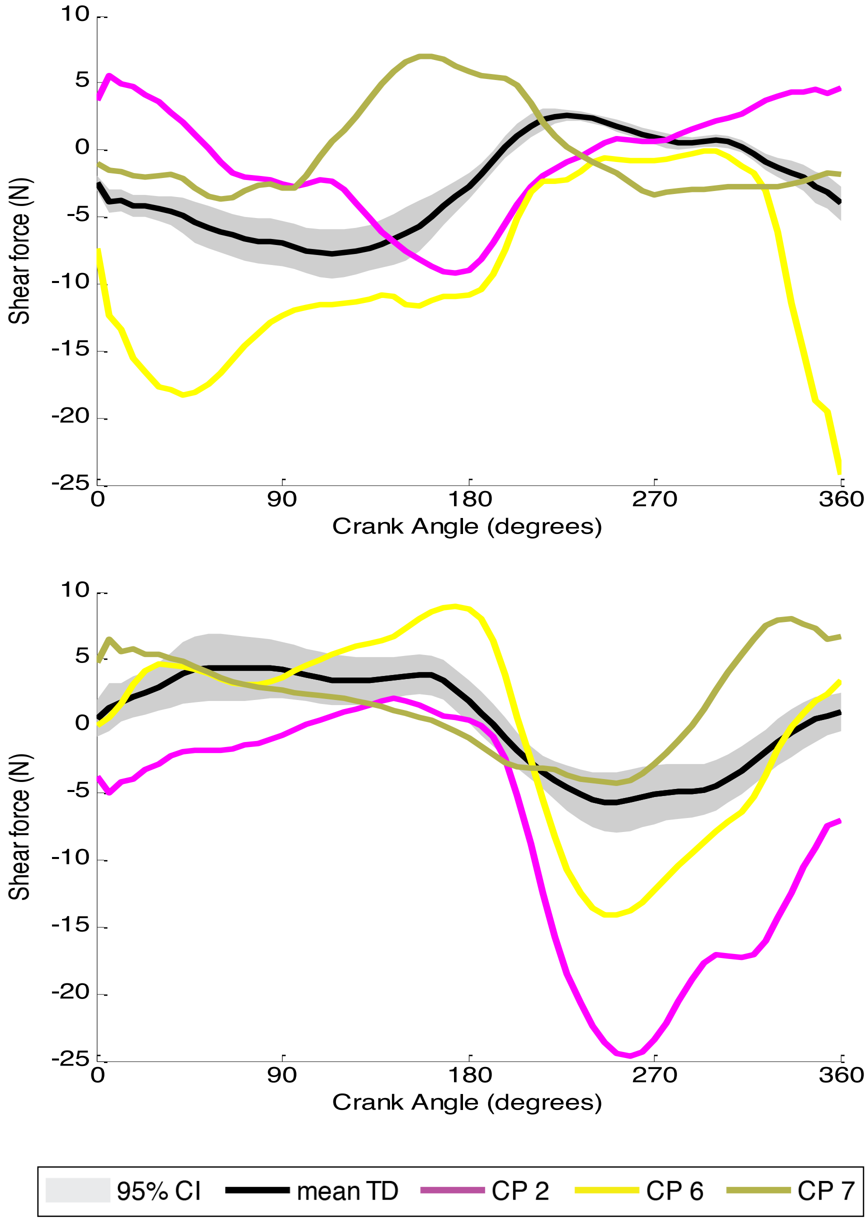


Figure A70: Shear forces at 60 RPM produced by right (top) and left (bottom) leg.

APPENDIX W: PEDAL FORCES AT 70 RPM IN STUDY 2

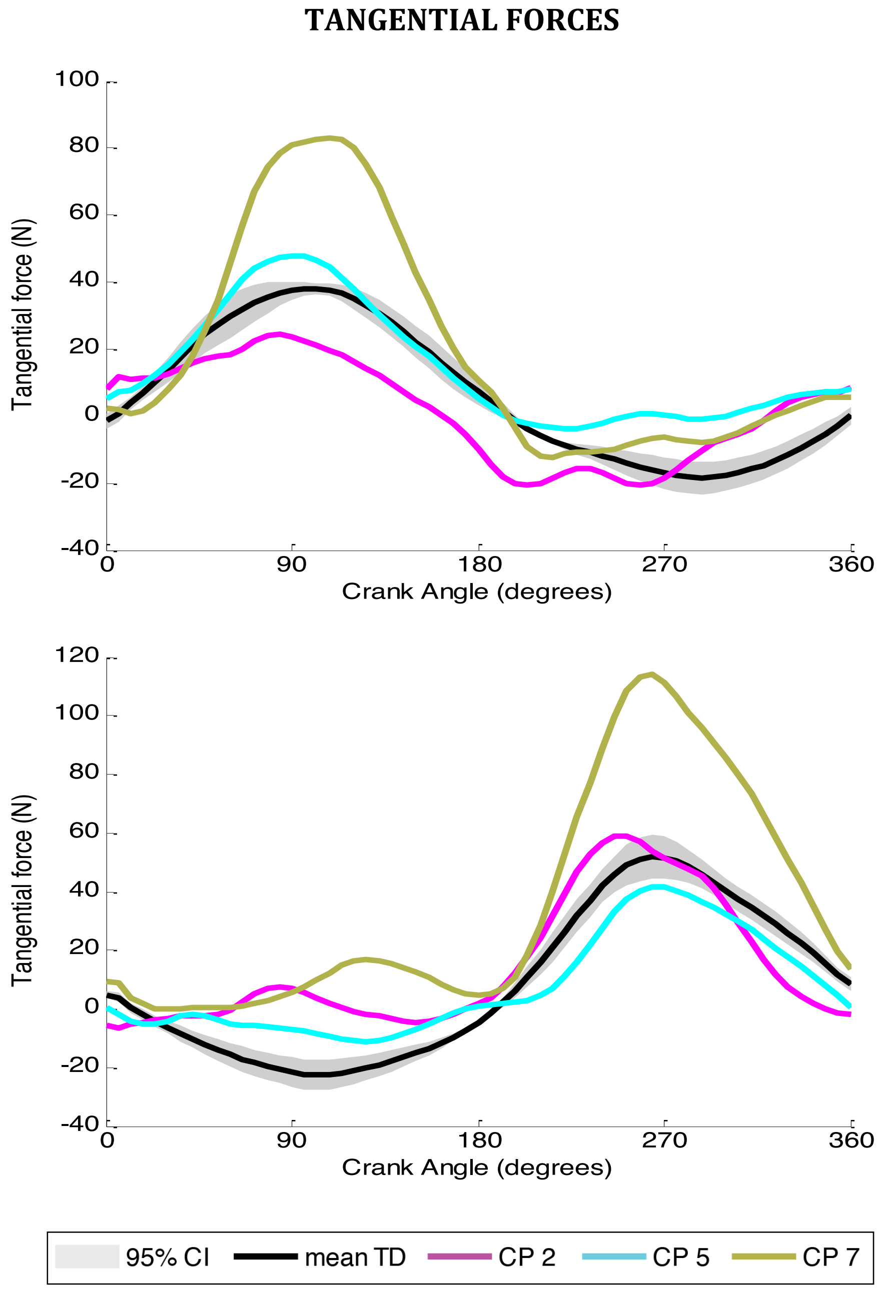


Figure A71: Tangential forces at 70 RPM produced by right (top) and left (bottom) leg.

NORMAL FORCES

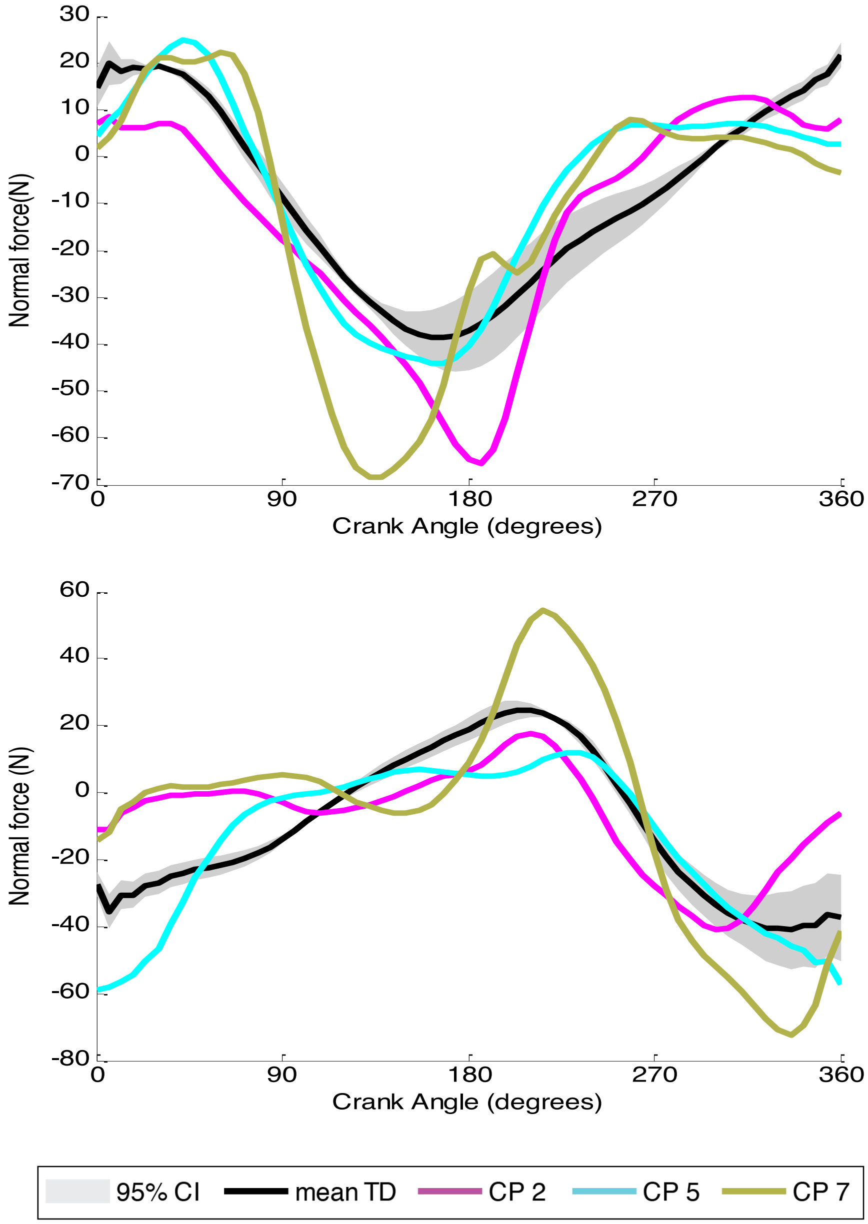


Figure A72: Normal forces at 70 RPM produced by right (top) and left (bottom) leg.

SHEAR FORCES

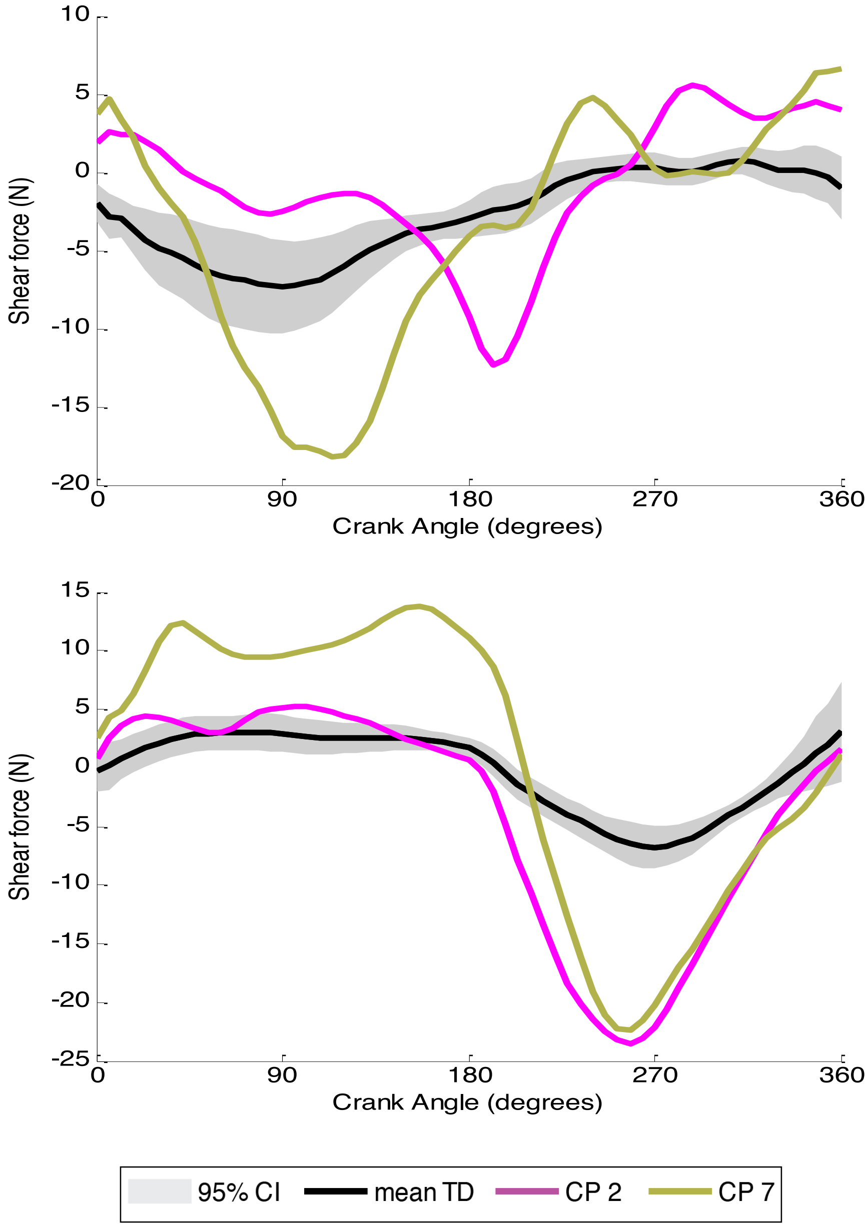


Figure A73: Shear forces at 70 RPM produced by right (top) and left (bottom) leg.

APPENDIX X: PEDAL FORCES AT 80 RPM IN STUDY 2

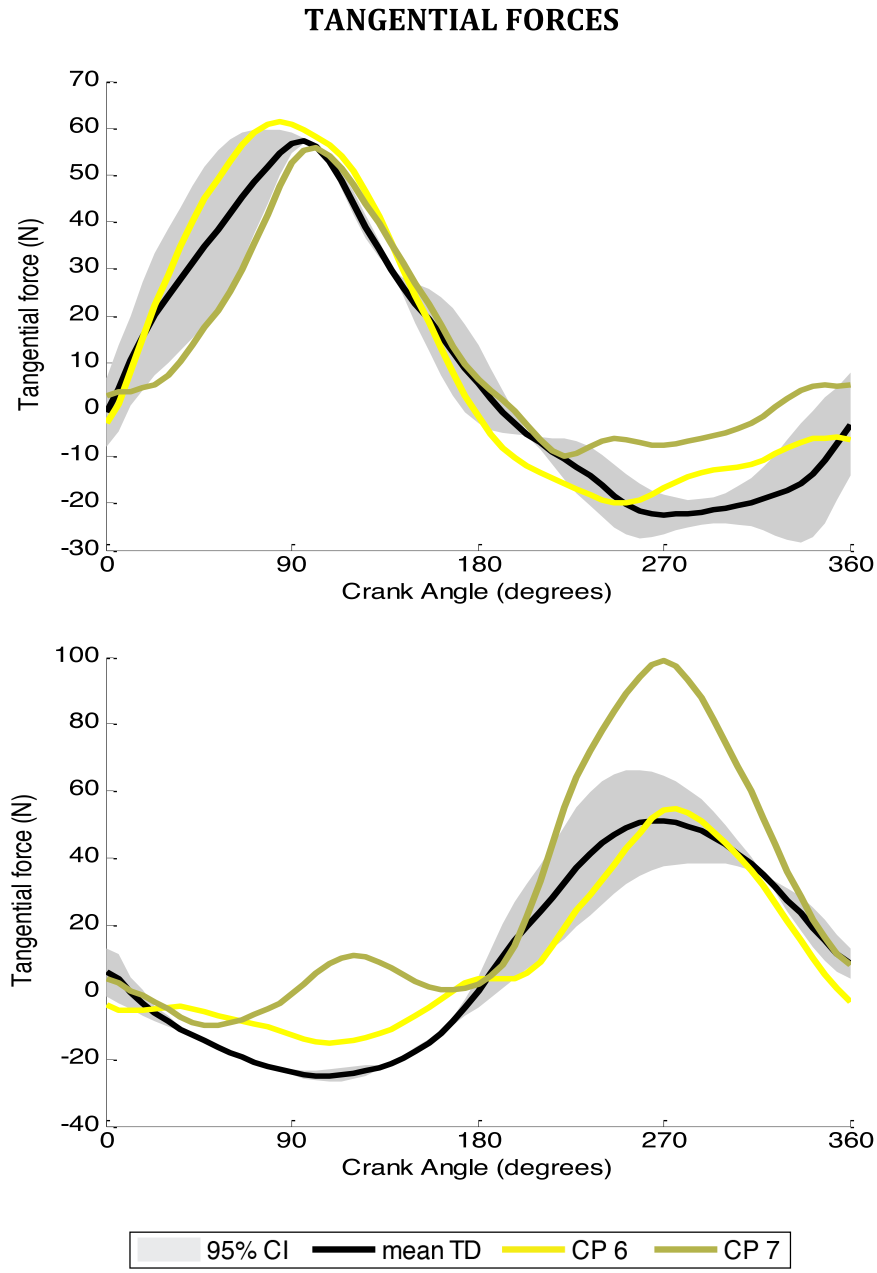


Figure A74: Tangential forces at 80 RPM produced by right (top) and left (bottom) leg.

NORMAL FORCES

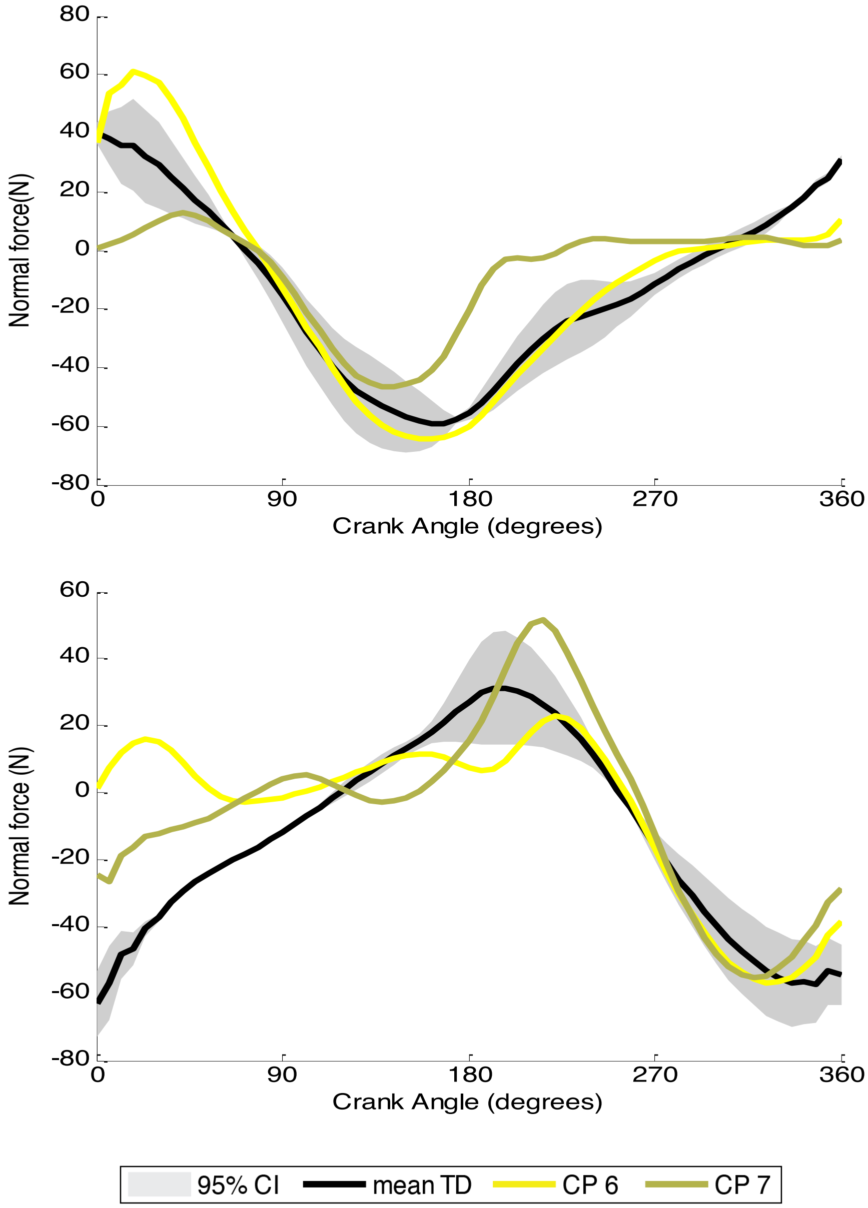


Figure A75: Normal forces at 80 RPM produced by right (top) and left (bottom) leg.

SHEAR FORCES

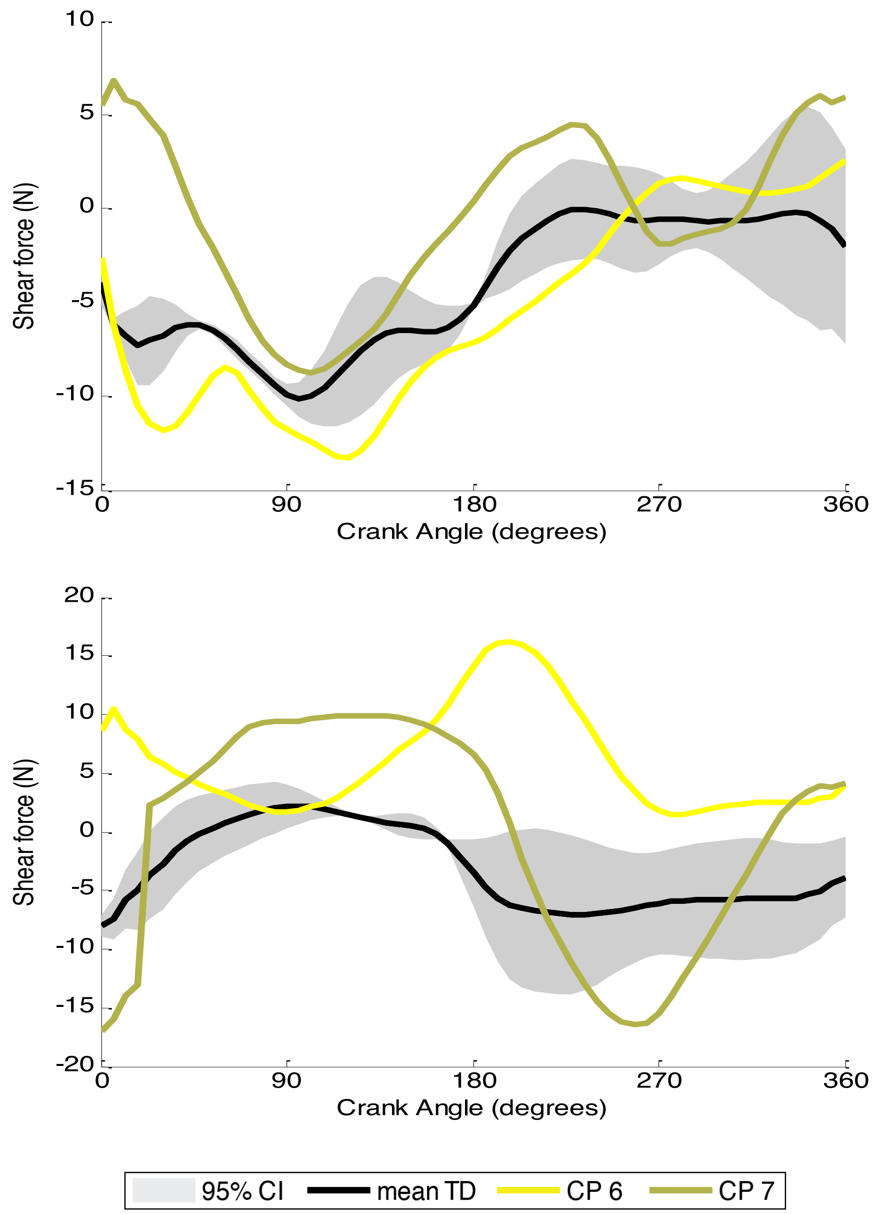


Figure A76: Shear forces at 80 RPM produced by right (top) and left (bottom) leg.

APPENDIX Y: JOINT TORQUES AT 30 RPM IN STUDY 2

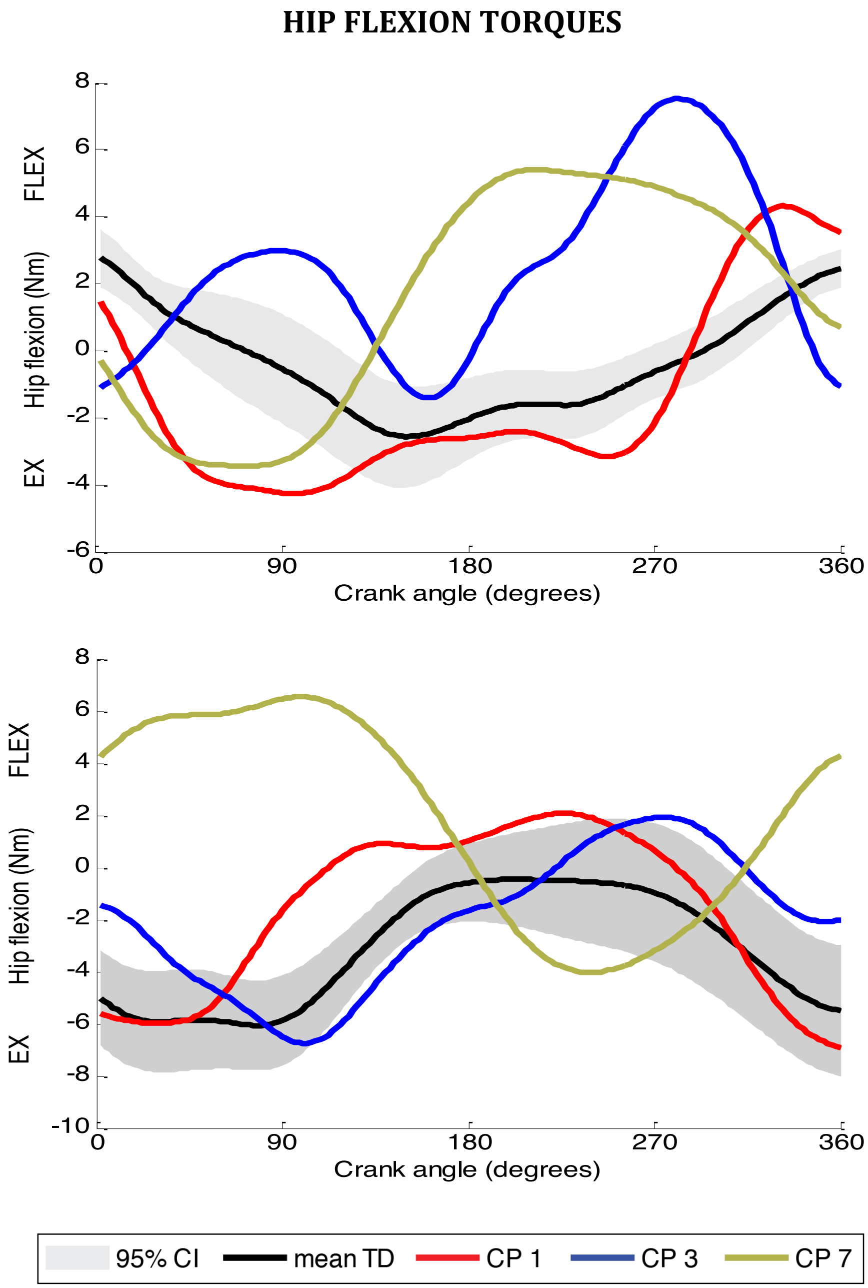


Figure A77: Hip flexion torques at 30 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION TORQUES

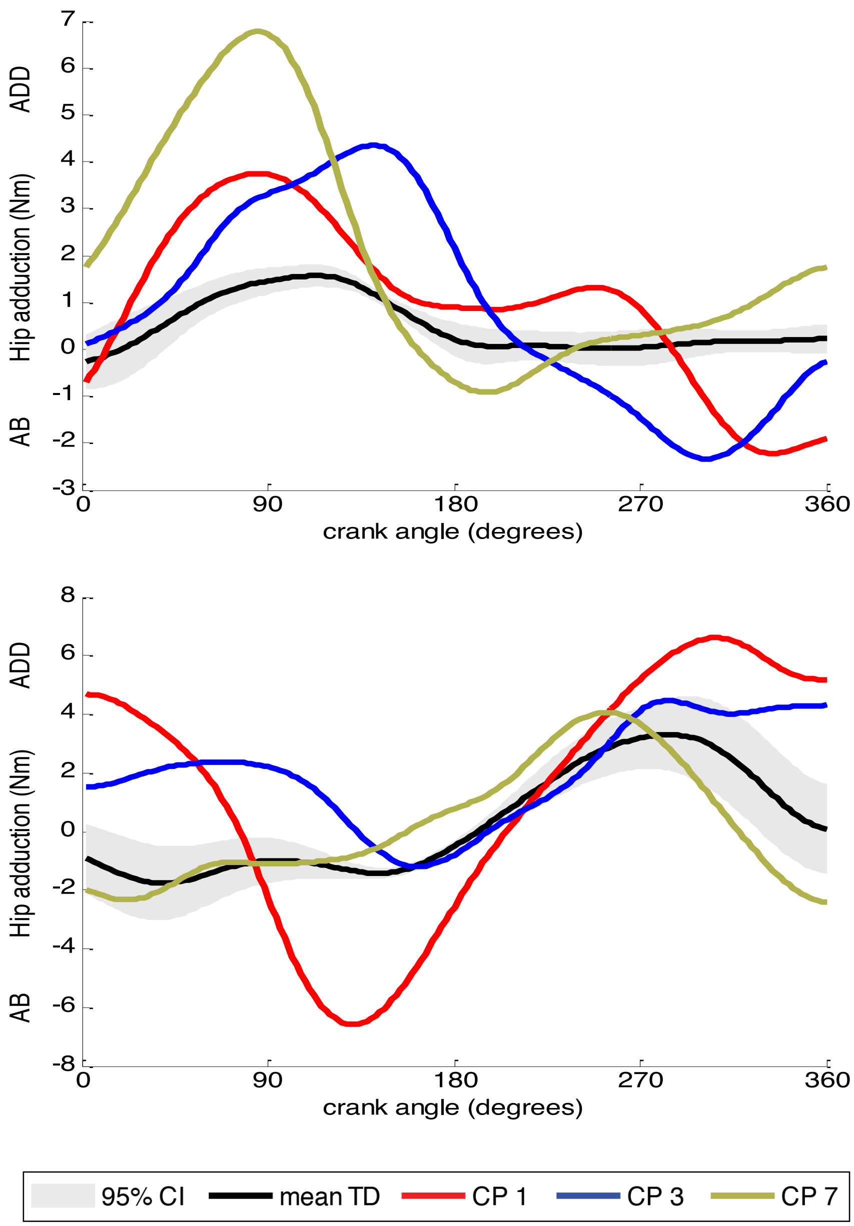


Figure A78: Hip adduction torques at 30 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION TORQUES

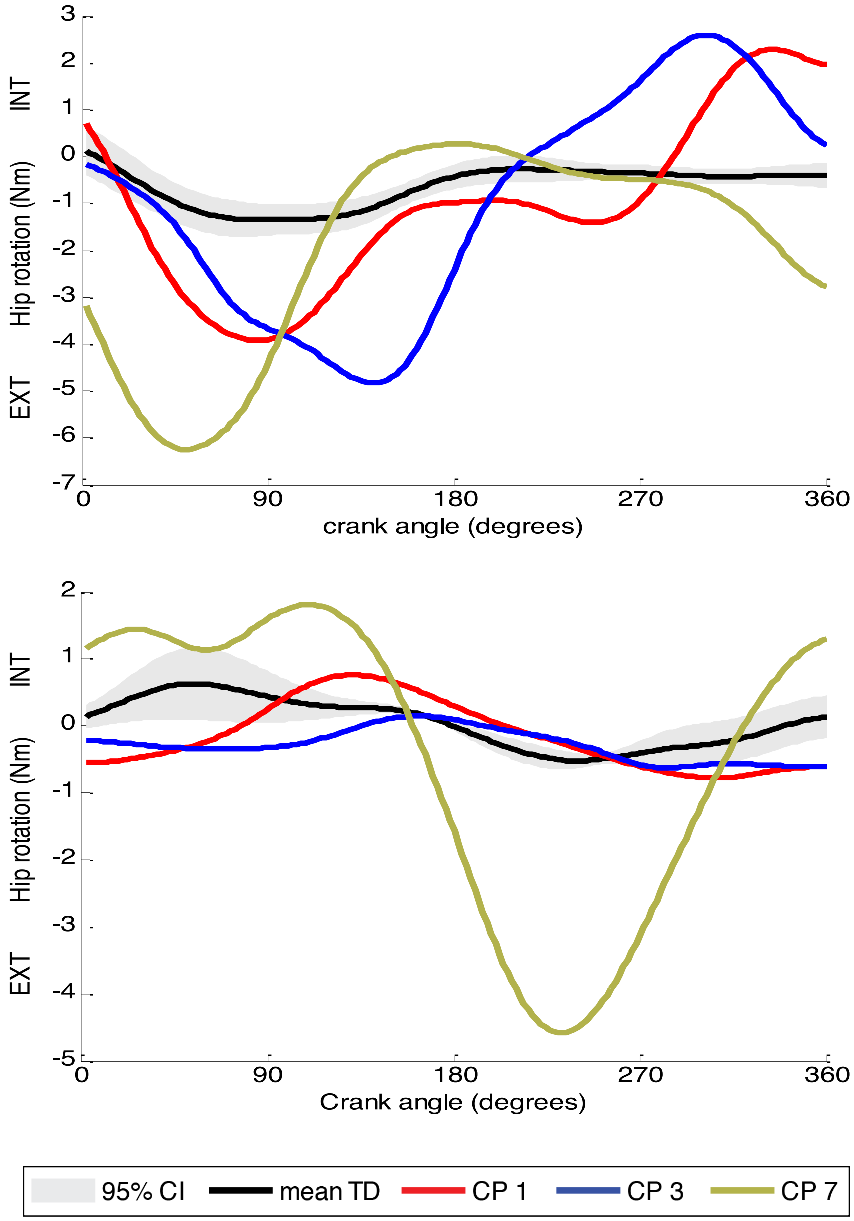


Figure A79: Hip rotation torques at 30 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION TORQUES

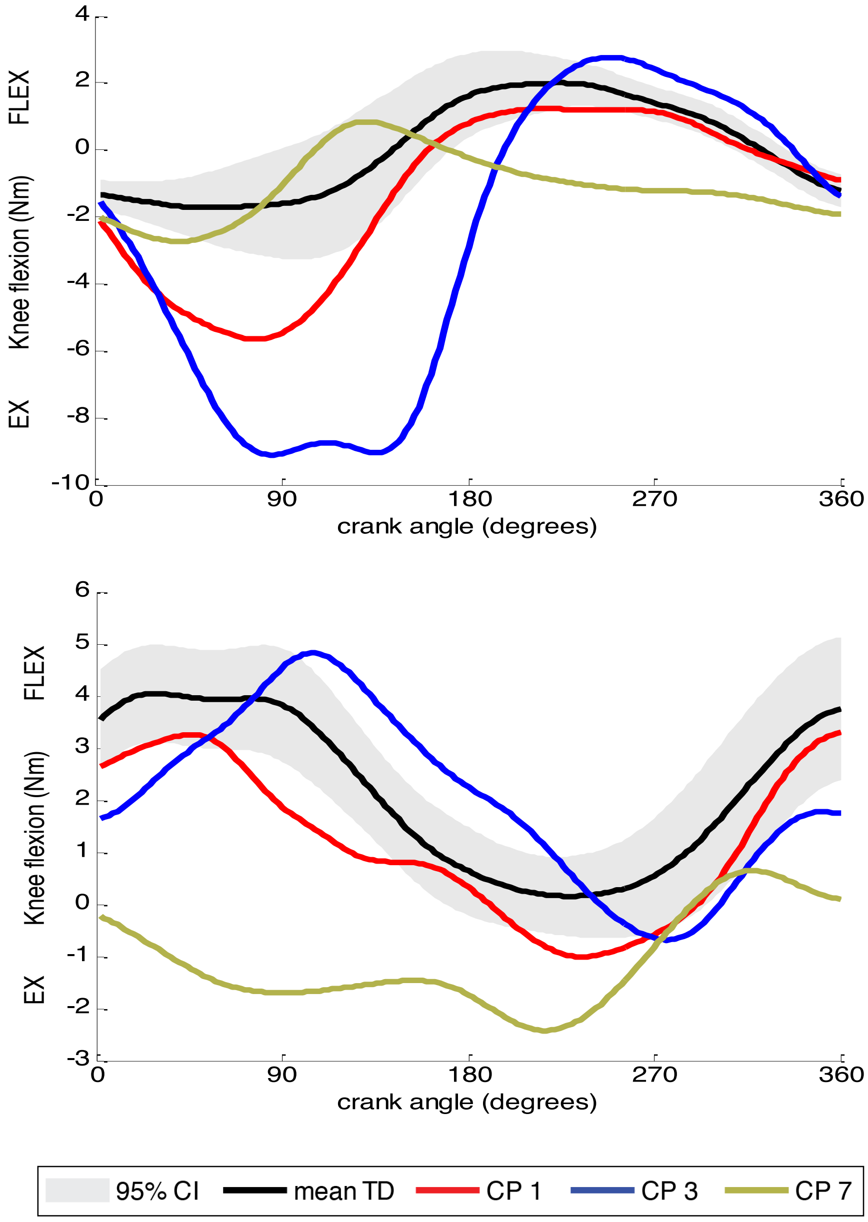


Figure A80: Knee flexion torques at 30 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION TORQUES

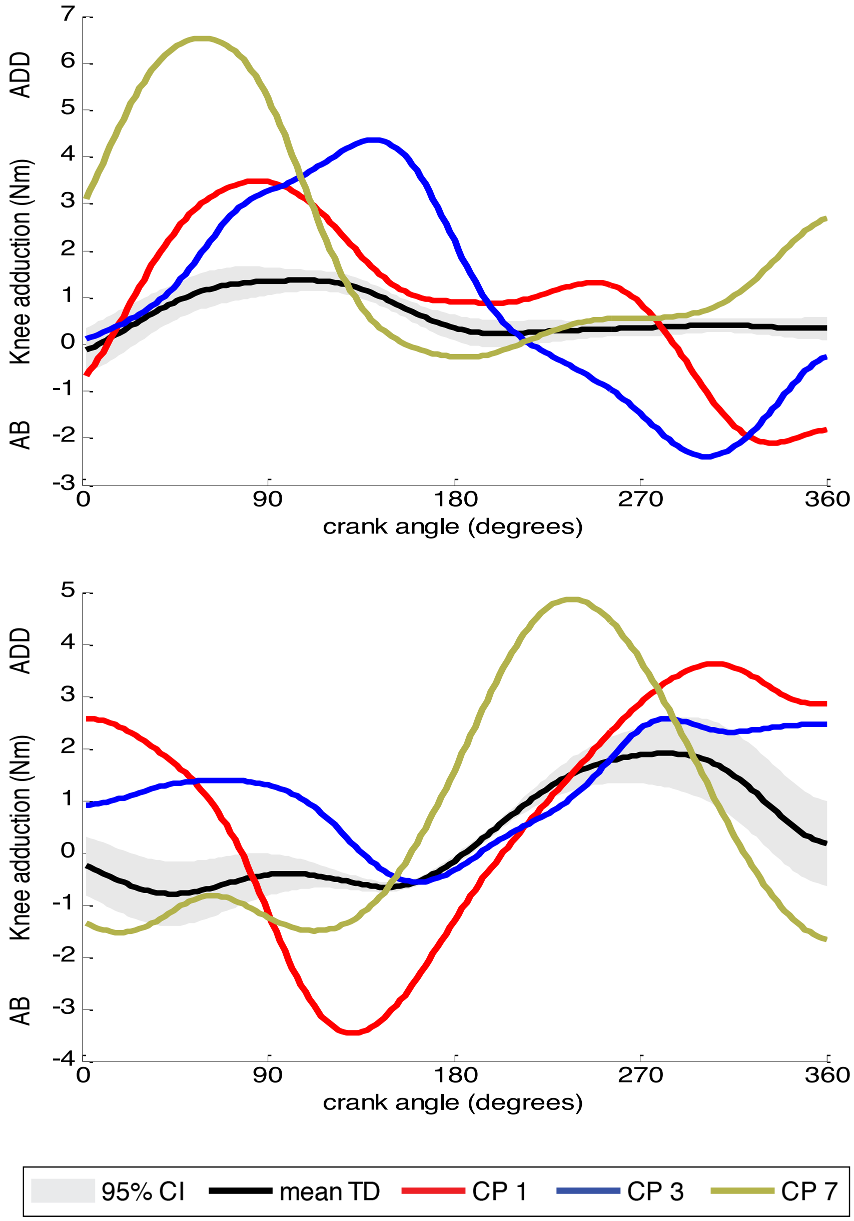


Figure A81: Knee adduction torques at 30 RPM produced by right (top) and left (bottom) leg.

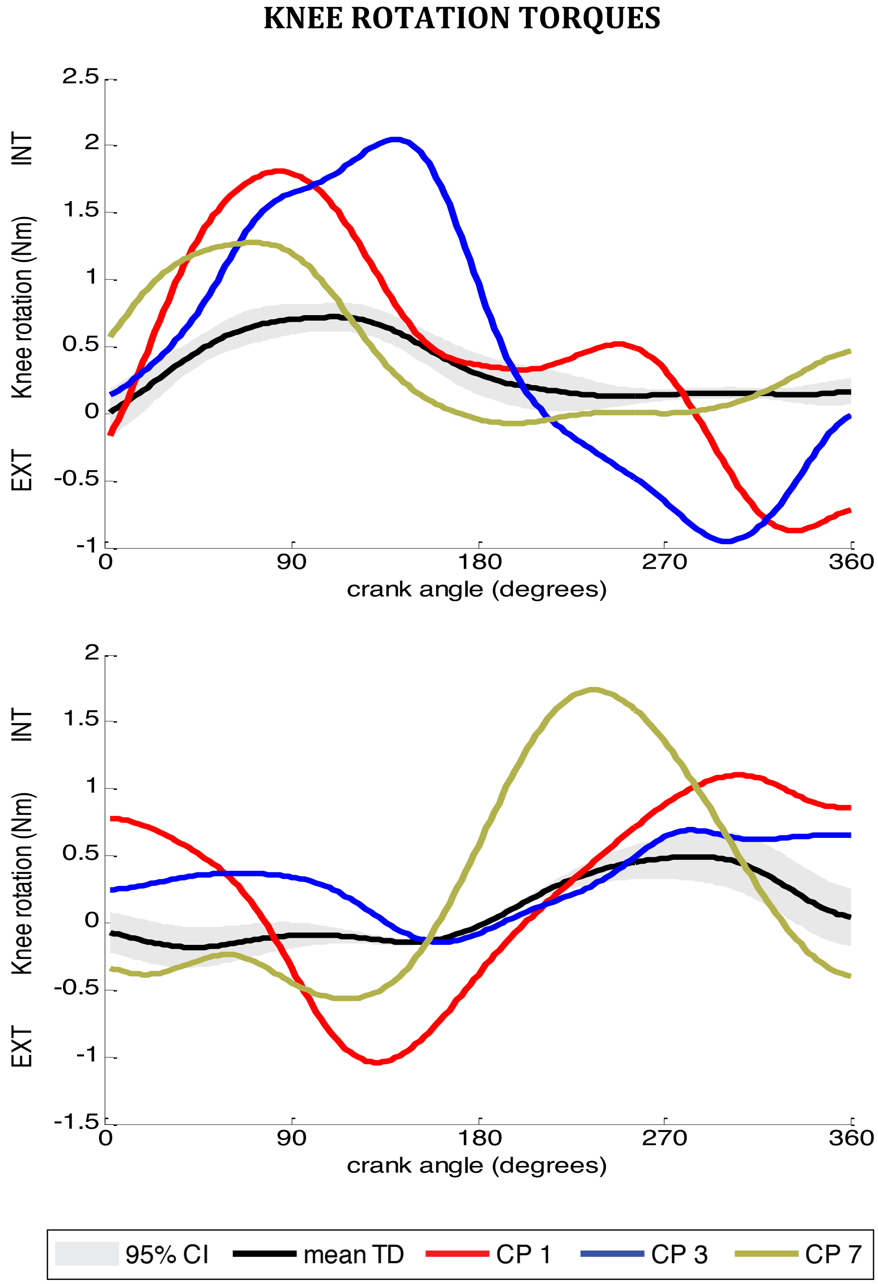


Figure A82: Knee rotation torques at 30 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION TORQUES

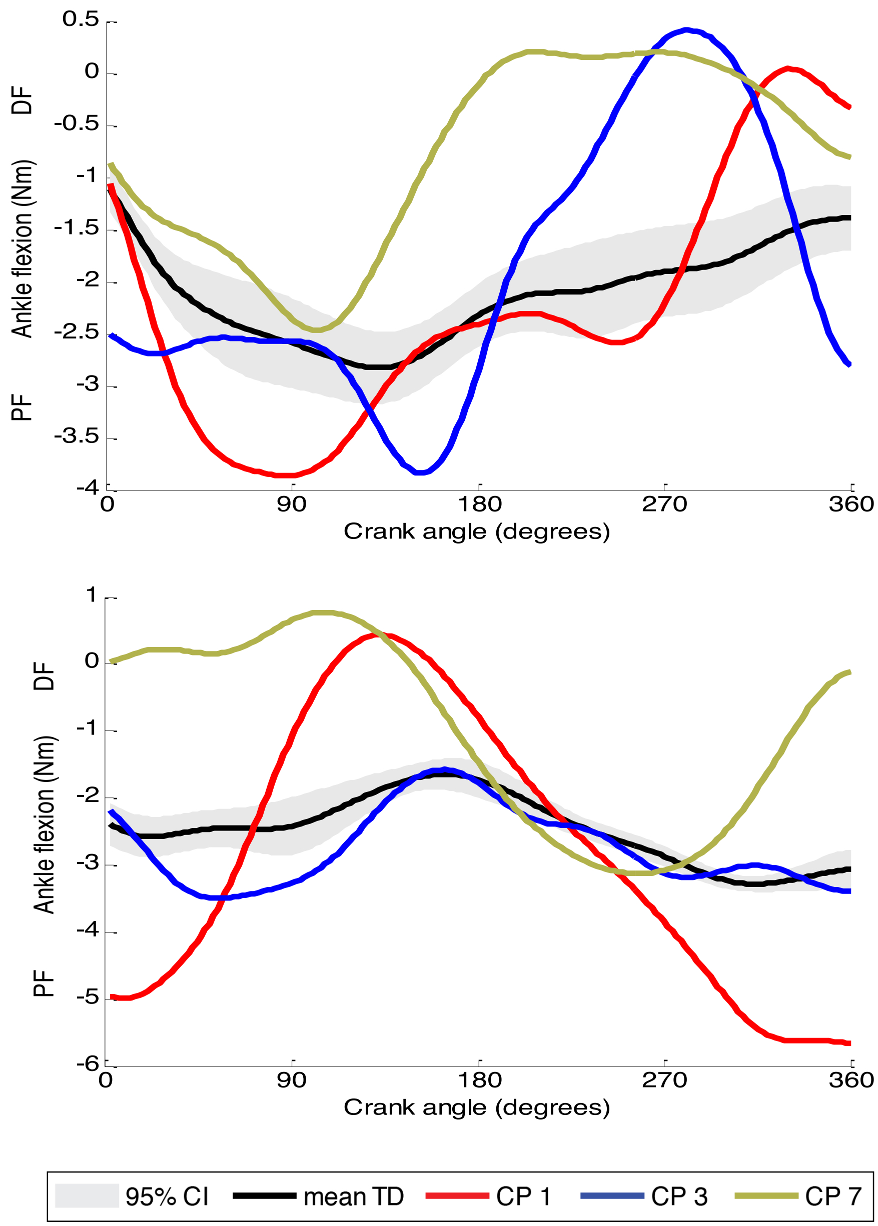


Figure A83: Ankle flexion torques at 30 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION TORQUES

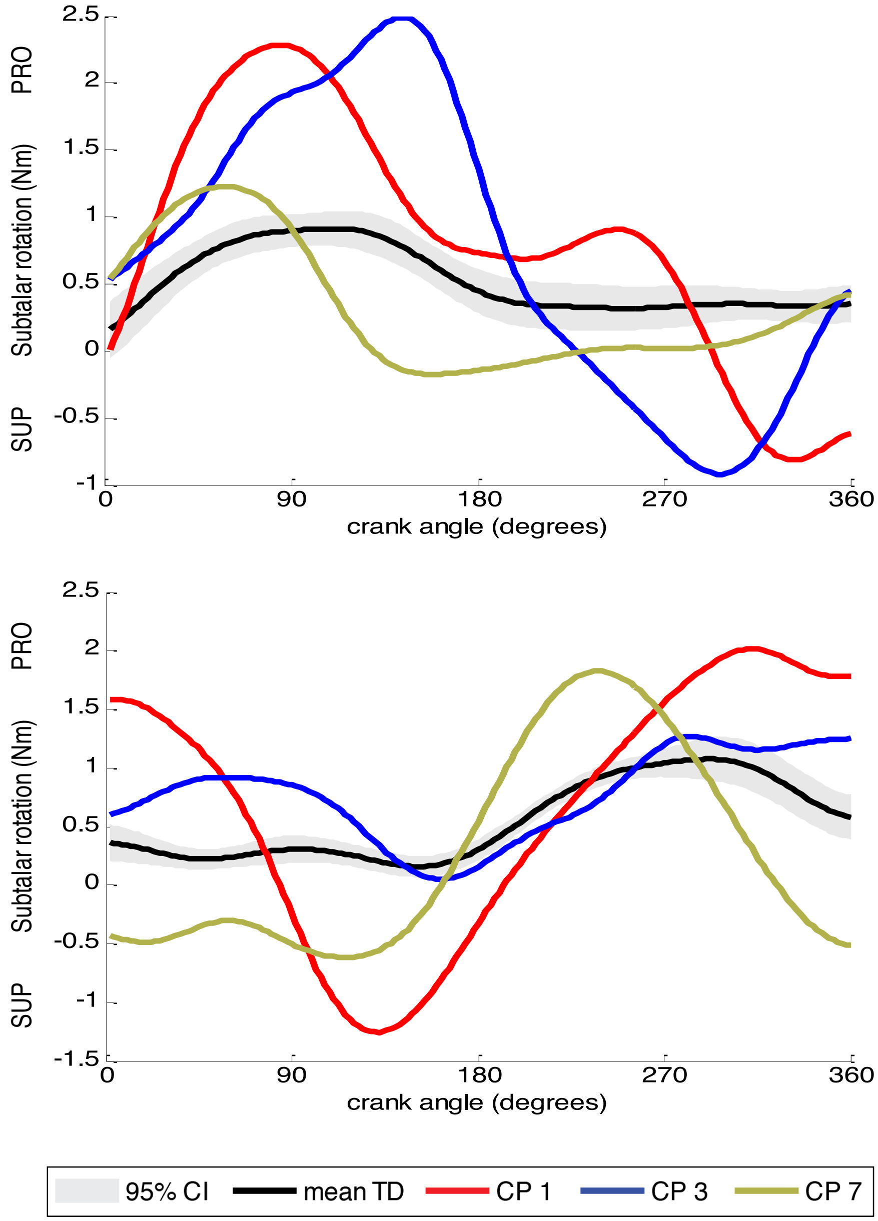


Figure A84: Subtalar rotation torques at 30 RPM produced by right (top) and left (bottom) leg.

APPENDIX Z: JOINT TORQUES AT 40 RPM IN STUDY 2

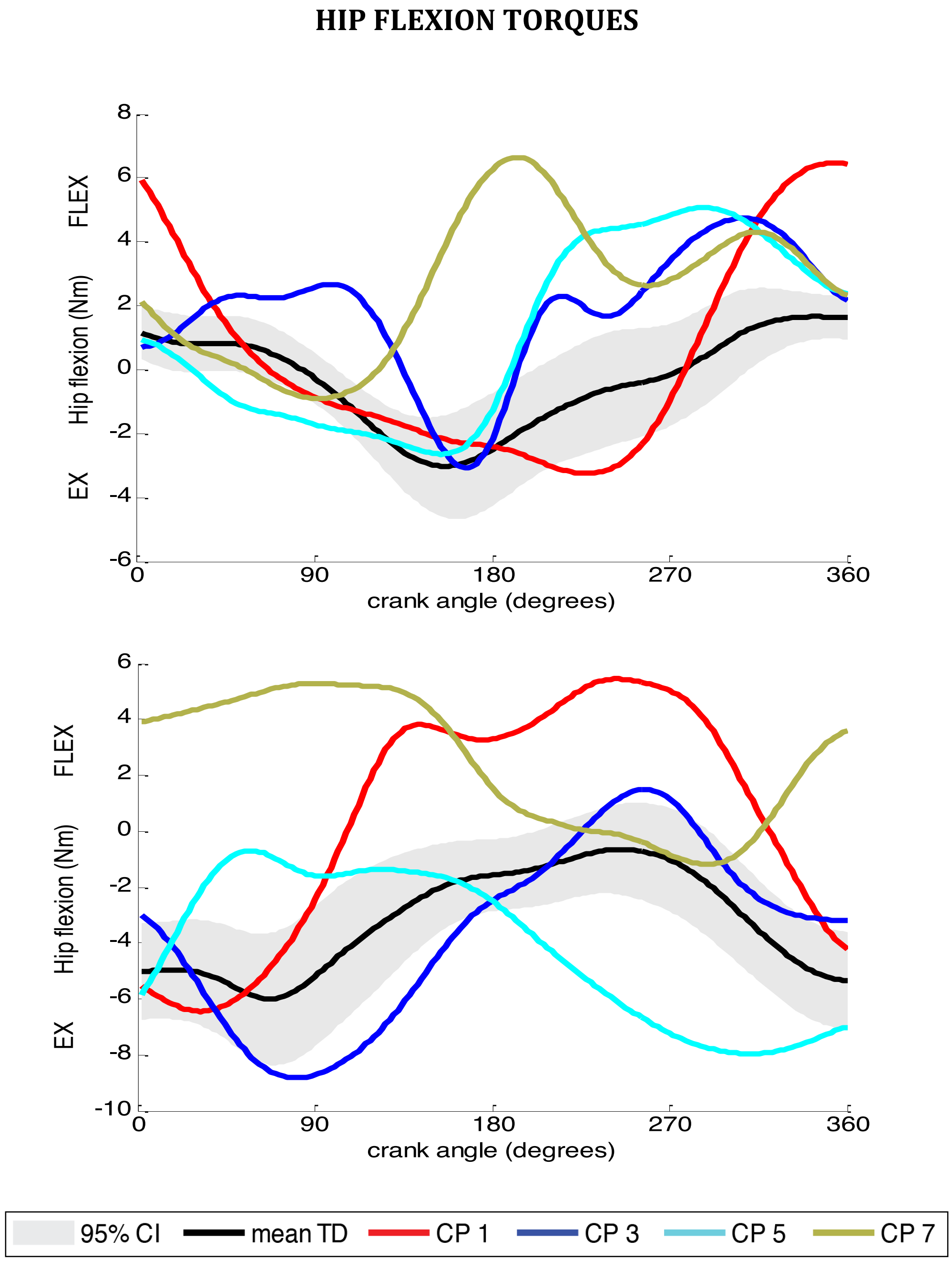


Figure A85: Hip flexion torques at 40 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION TORQUES

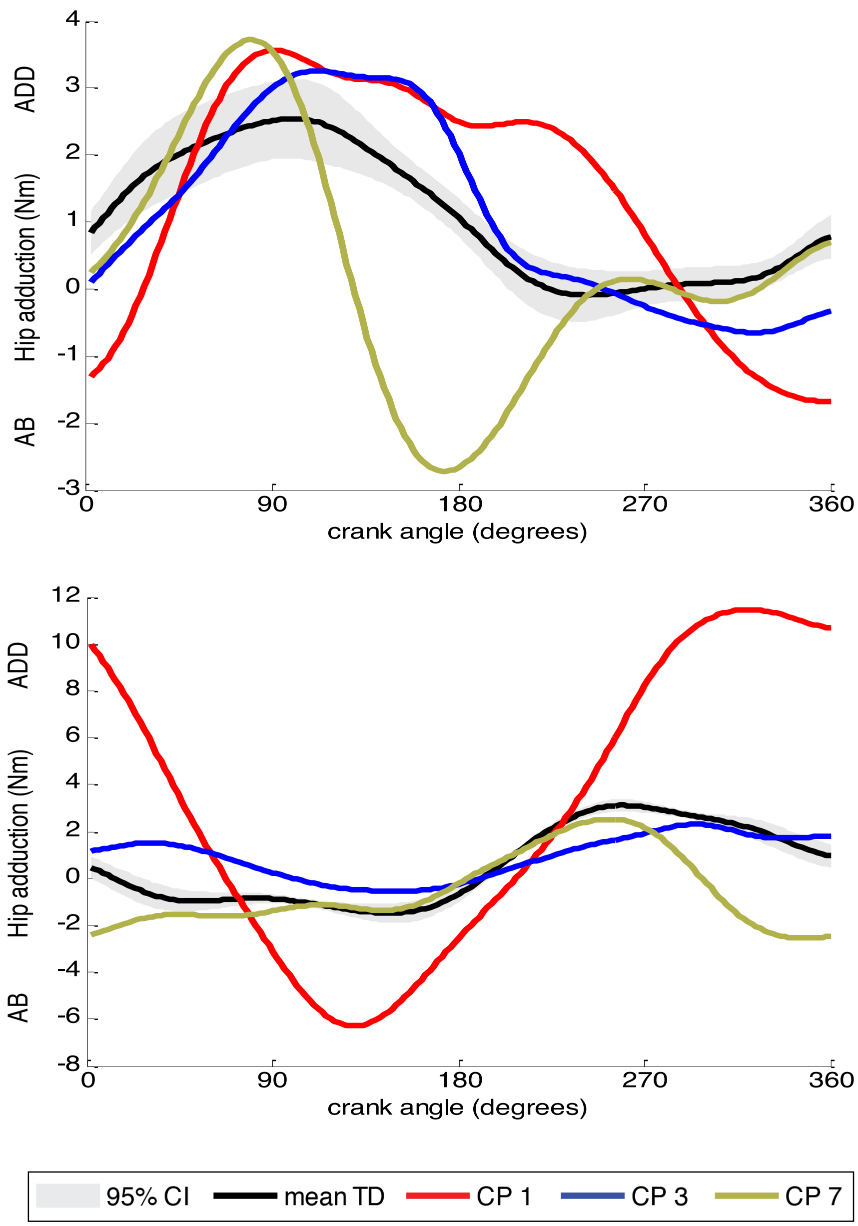


Figure A86: Hip adduction torques at 40 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION TORQUES

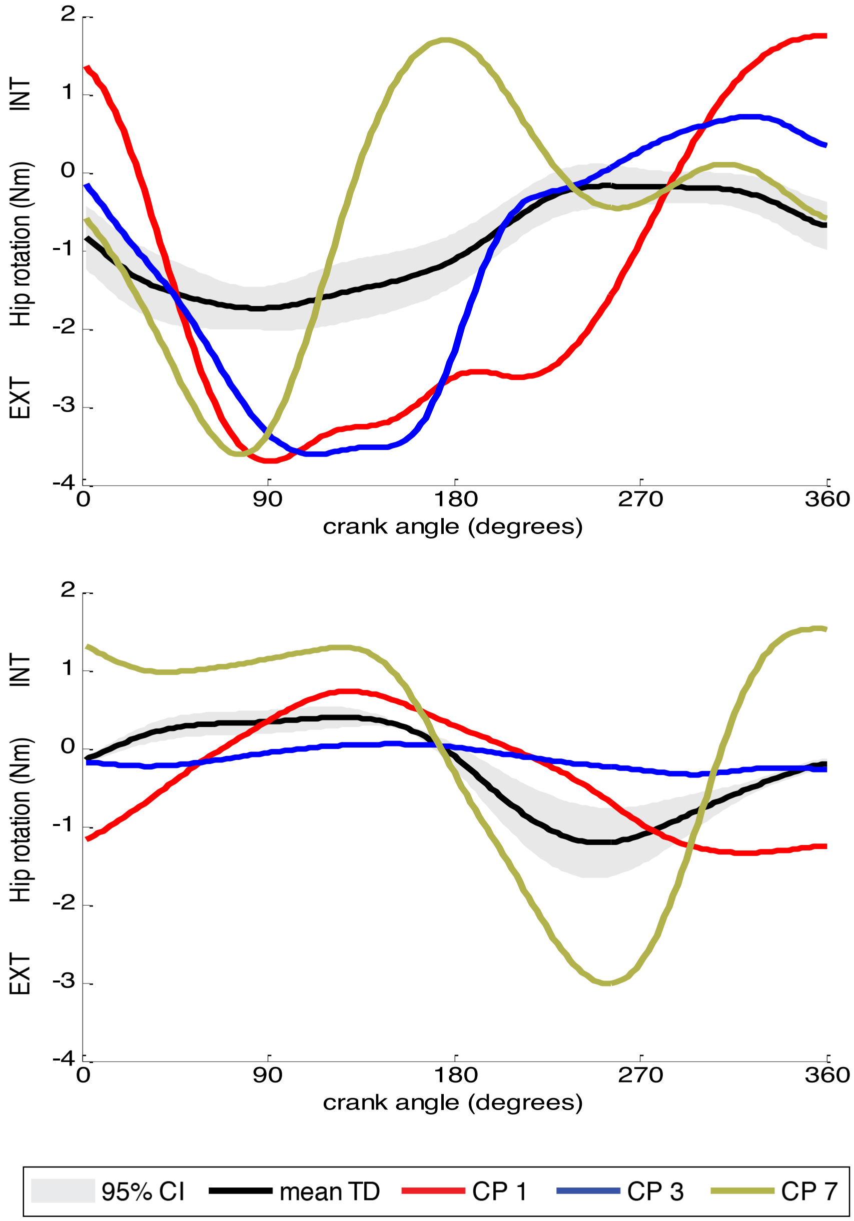


Figure A87: Hip rotation torques at 40 RPM produced by right (top) and left (bottom) leg.

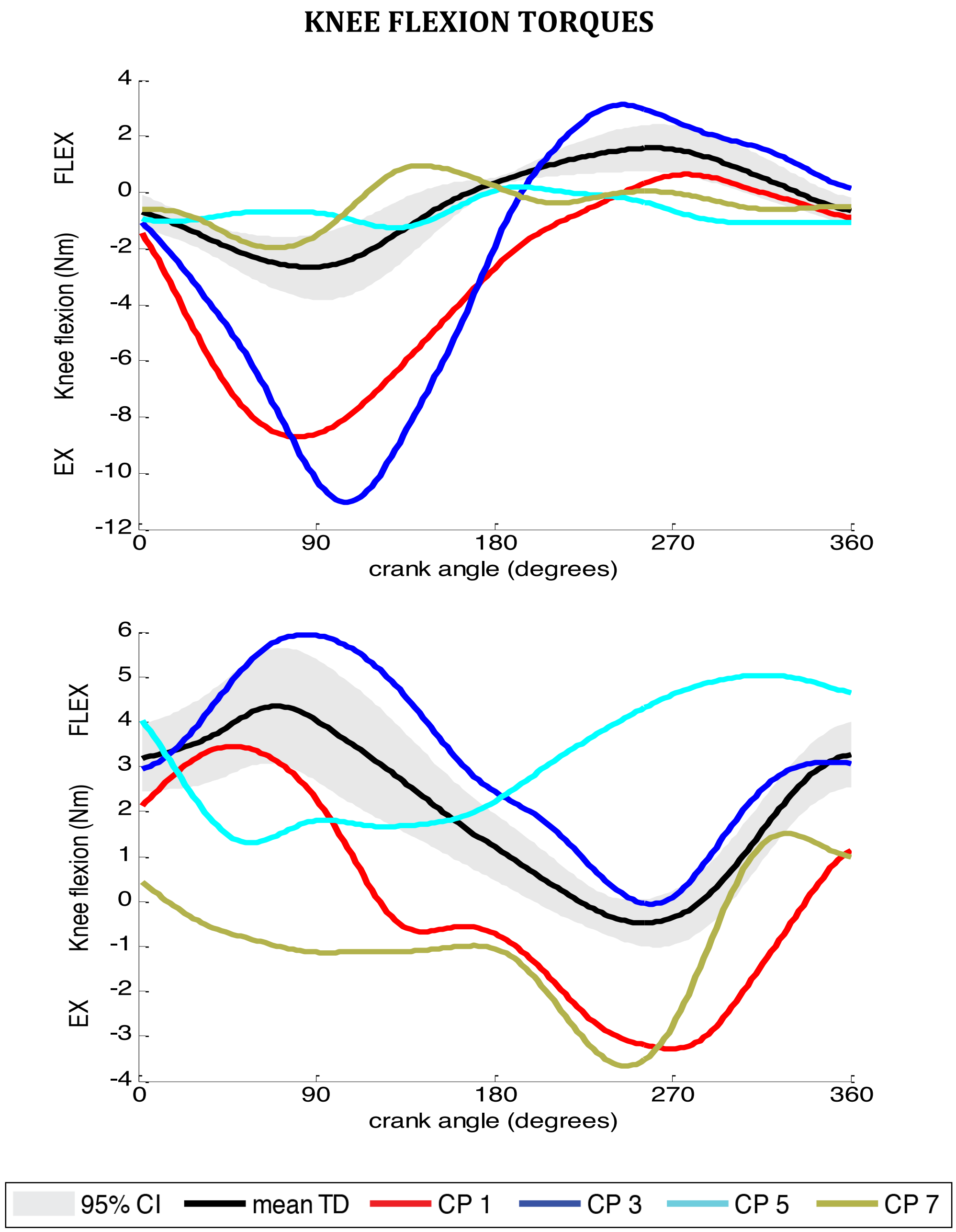


Figure A88: Knee flexion torques at 40 RPM produced by right (top) and left (bottom) leg.

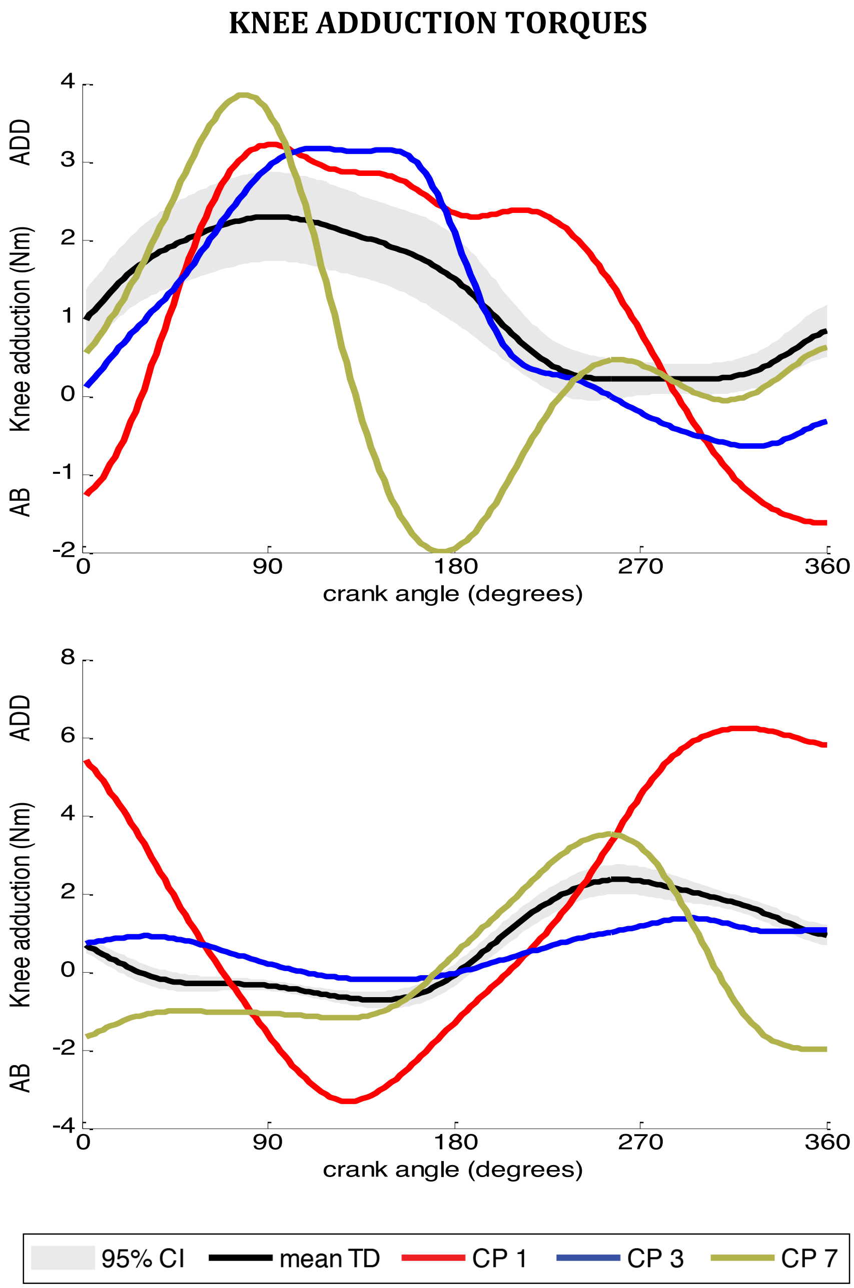


Figure A89: Knee adduction torques at 40 RPM produced by right (top) and left (bottom) leg.

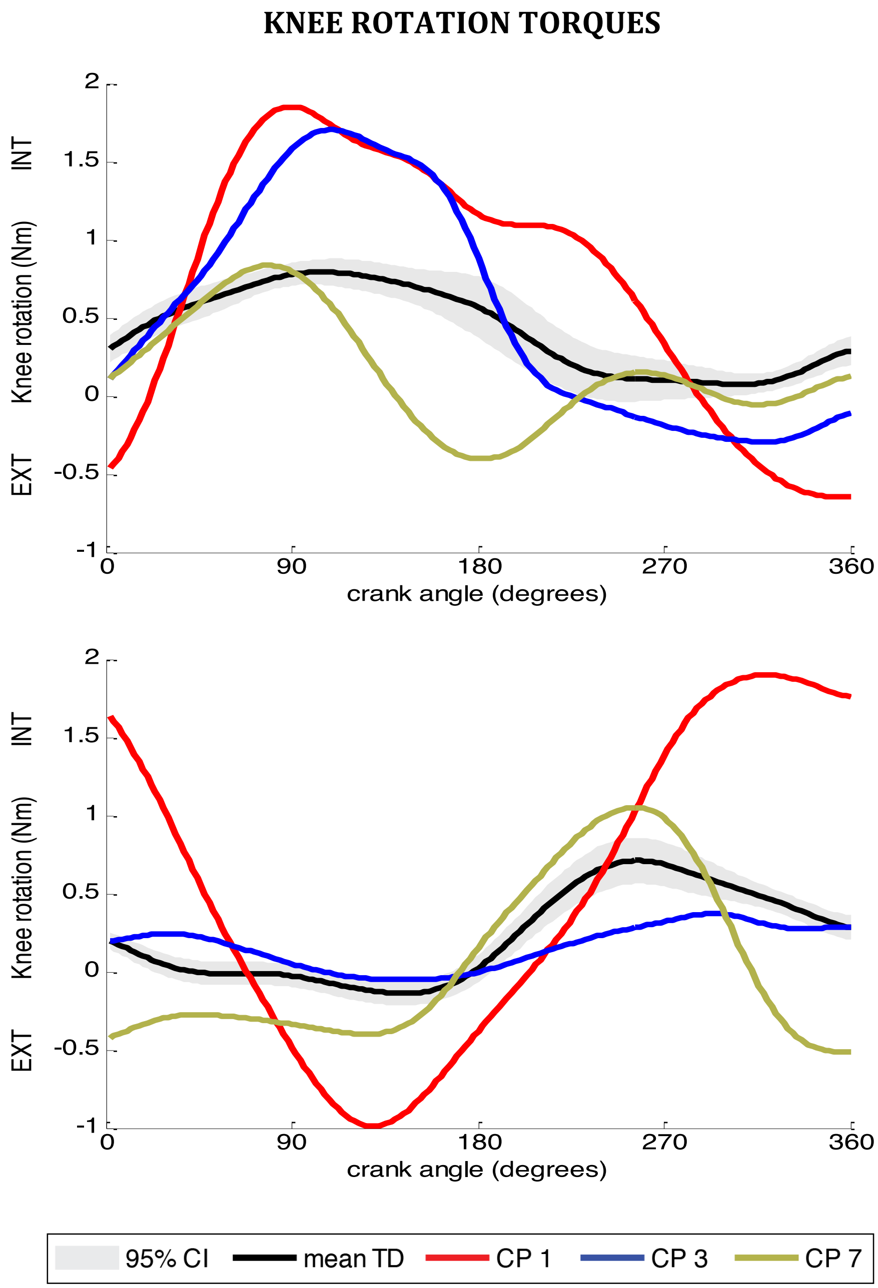


Figure A90: Knee rotation torques at 40 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION TORQUES

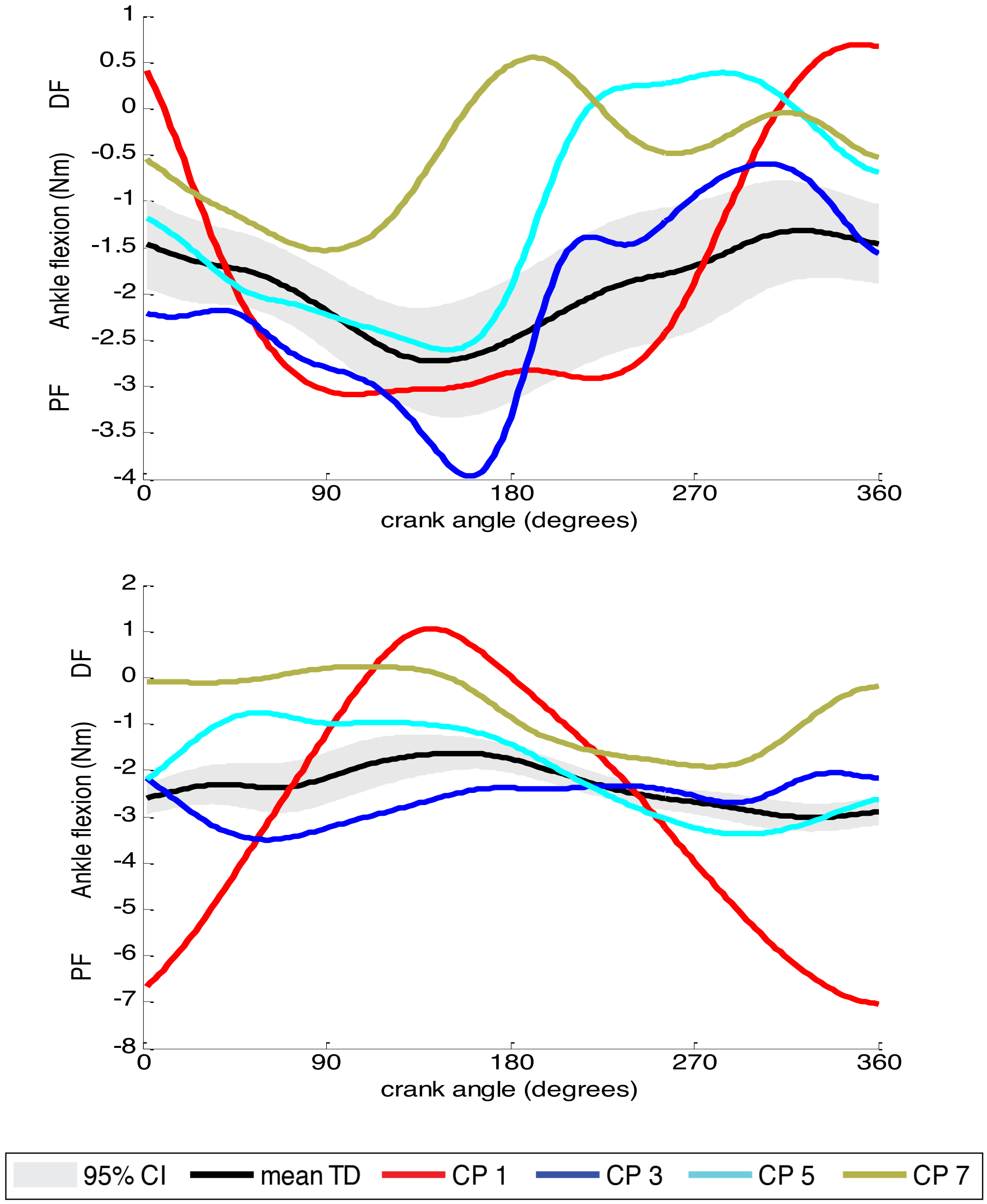


Figure A91: Ankle flexion torques at 40 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION TORQUES

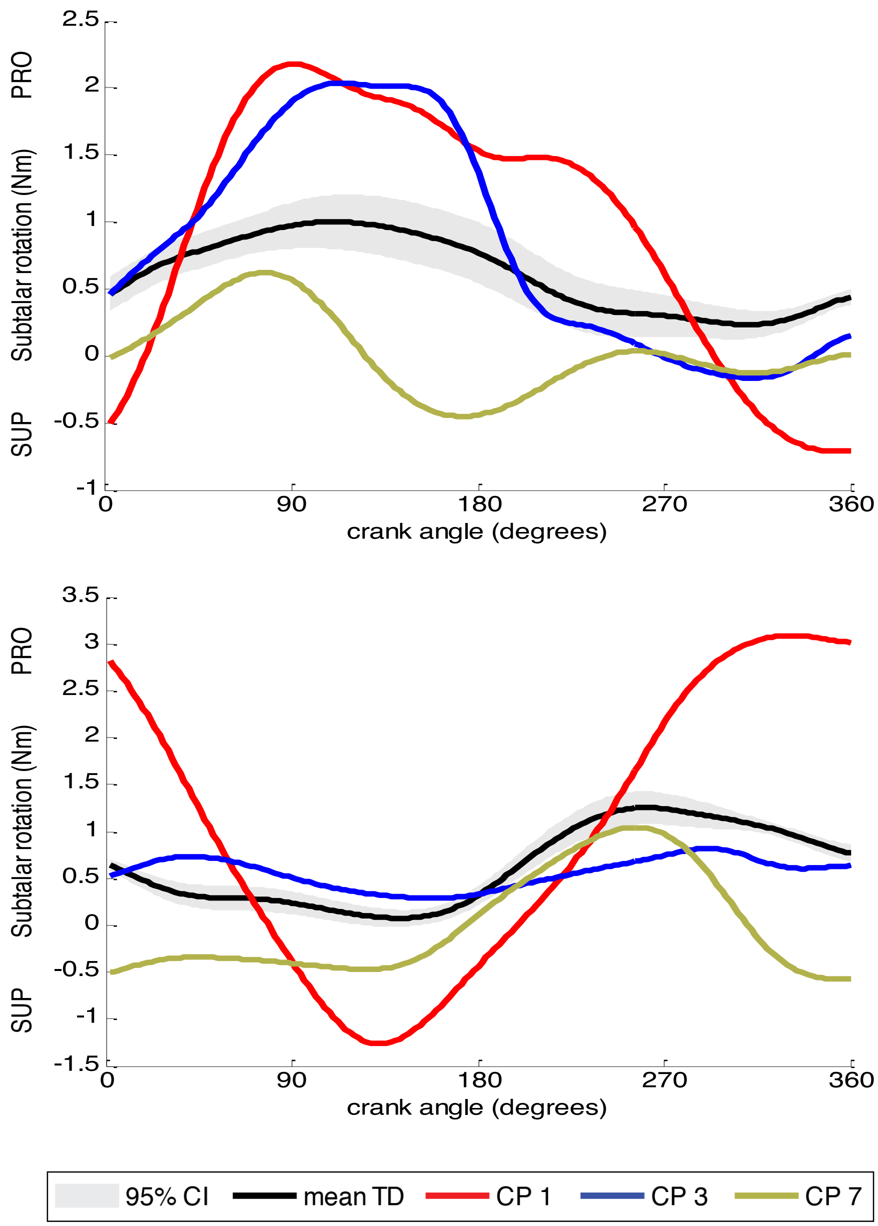


Figure A92: Subtalar rotation torques at 40 RPM produced by right (top) and left (bottom) leg.

Appendix AA: Joint torques at 50 RPM in Study 2

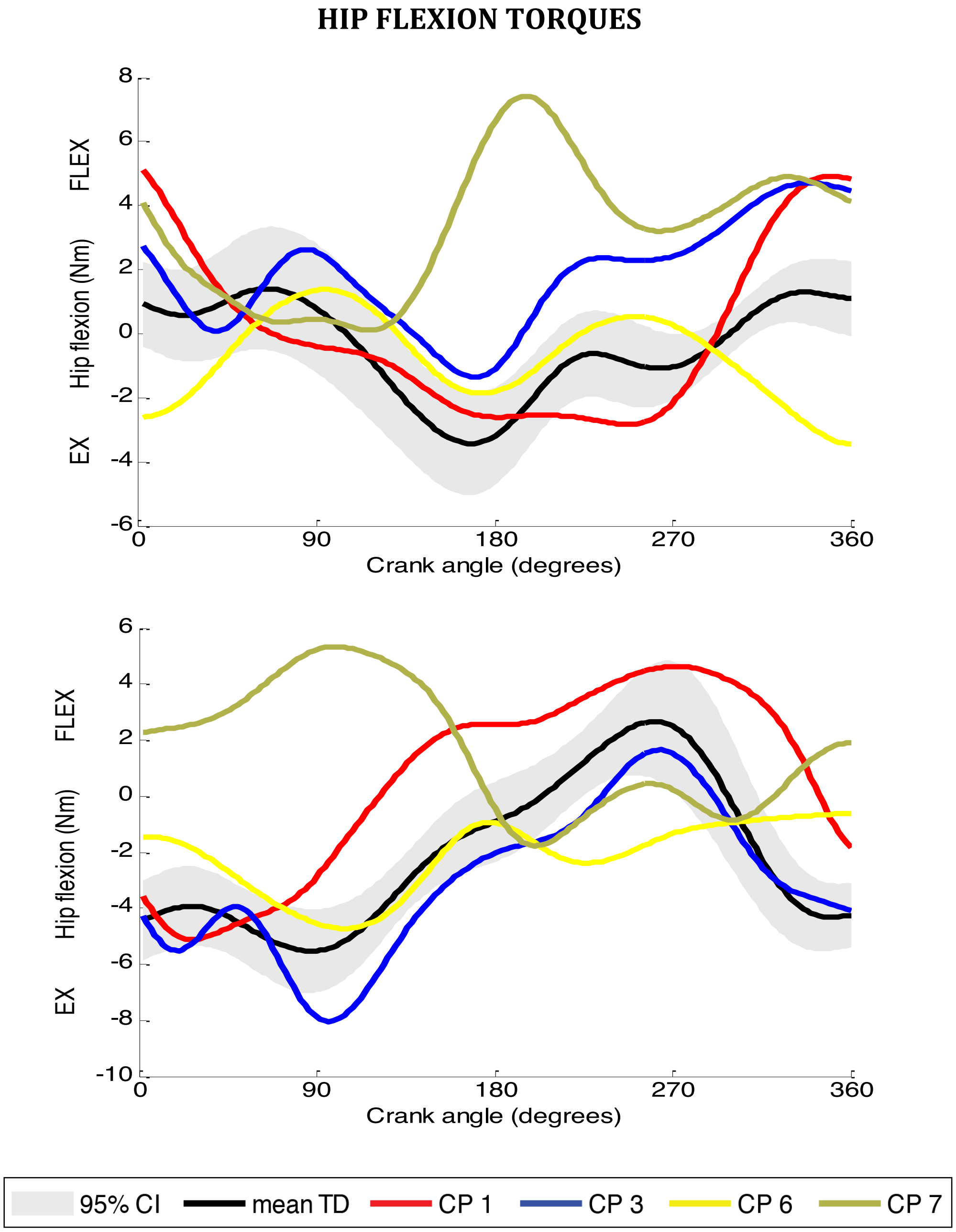


Figure A93: Hip flexion torques at 50 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION TORQUES

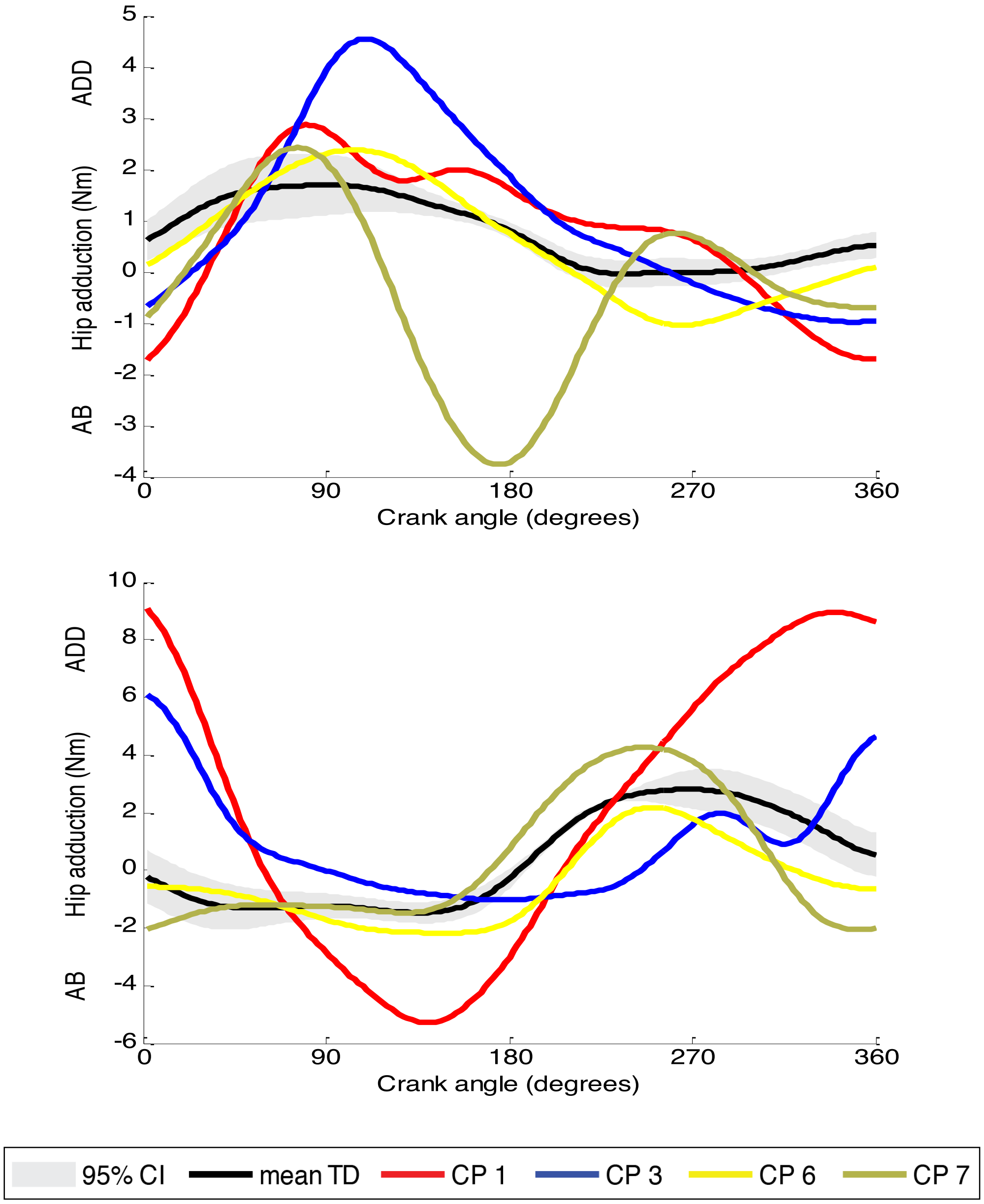


Figure A94: Hip adduction torques at 50 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION TORQUES

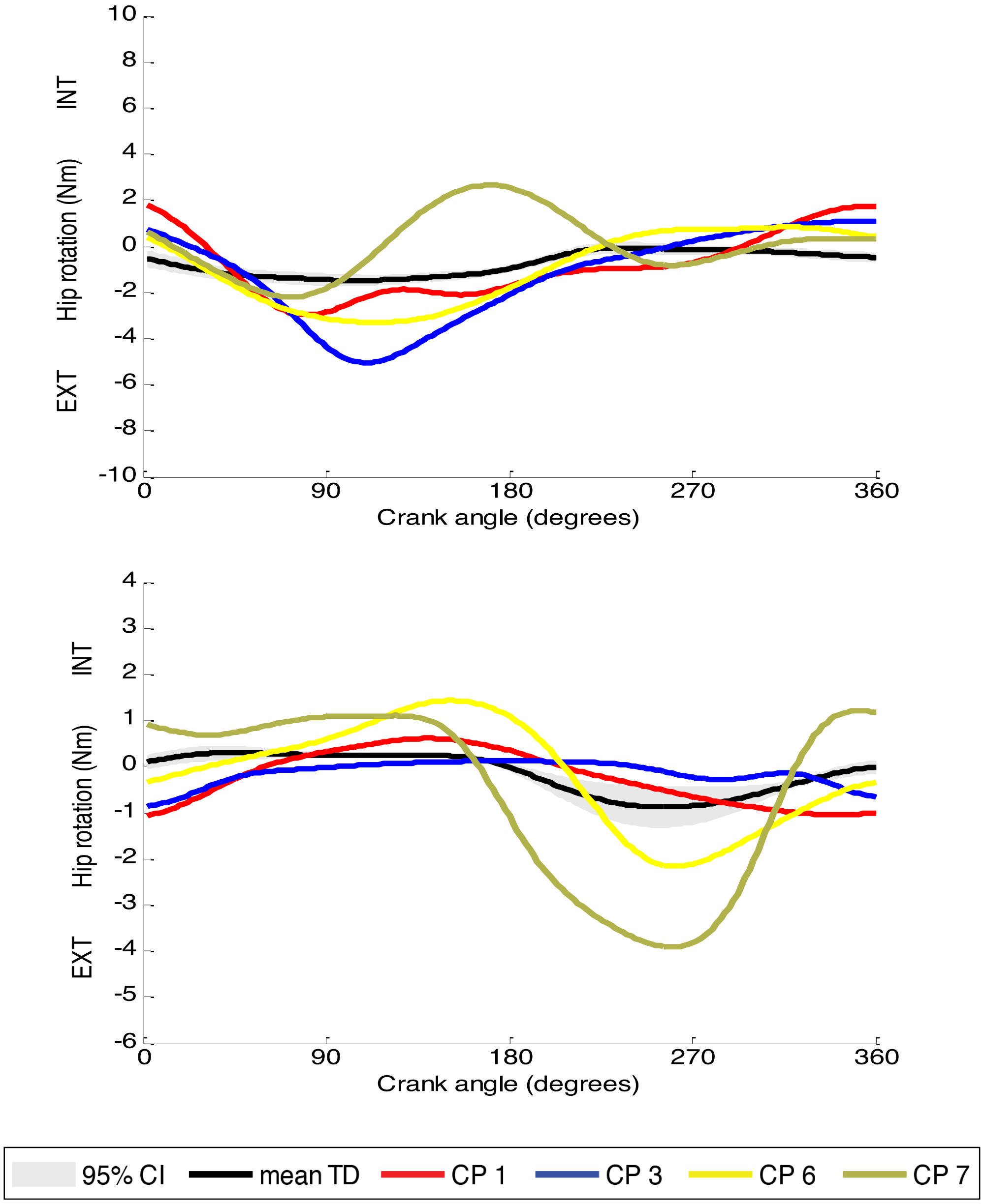


Figure A95: Hip rotation torques at 50 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION TORQUES

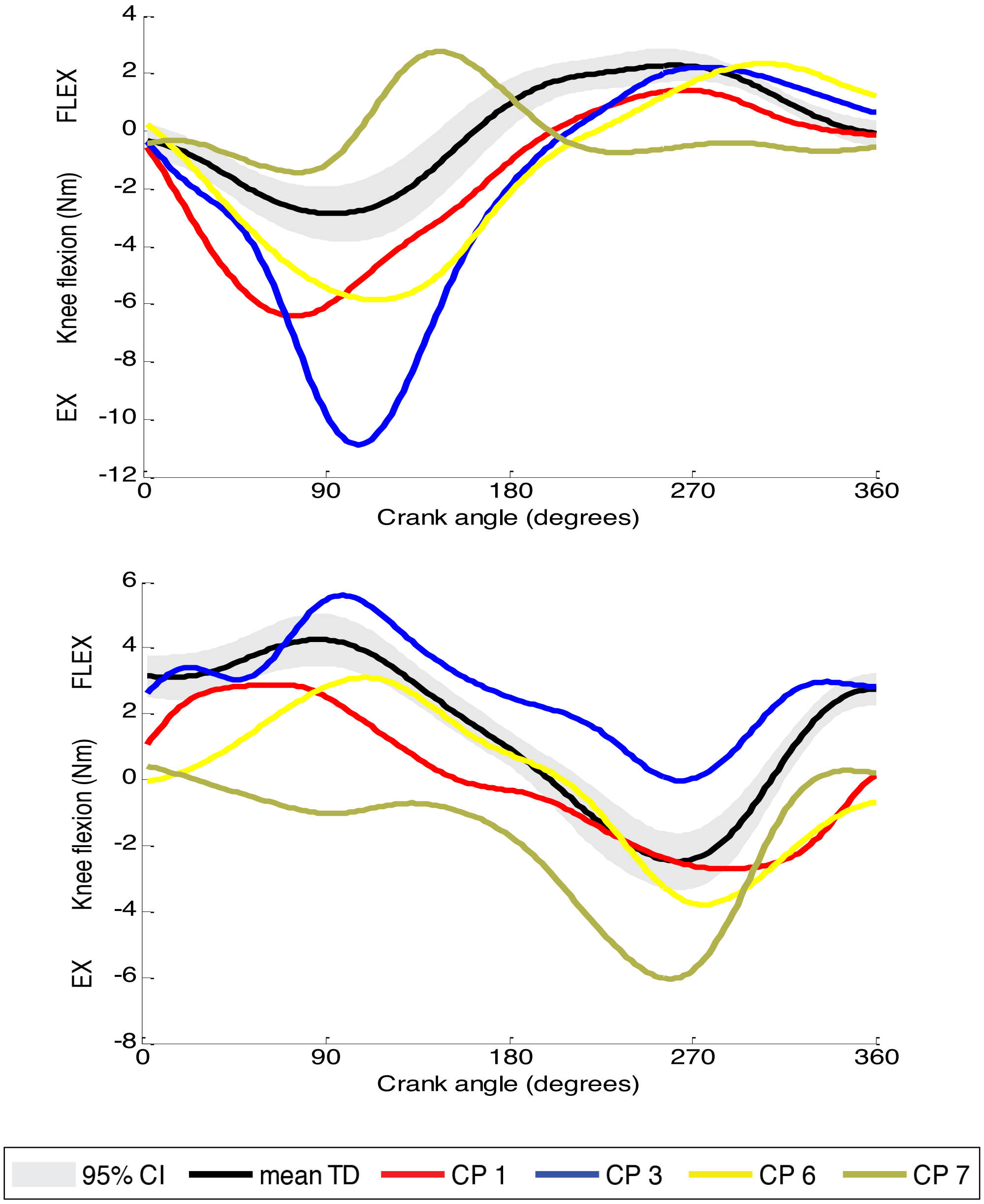


Figure A96: Knee flexion torques at 50 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION TORQUES

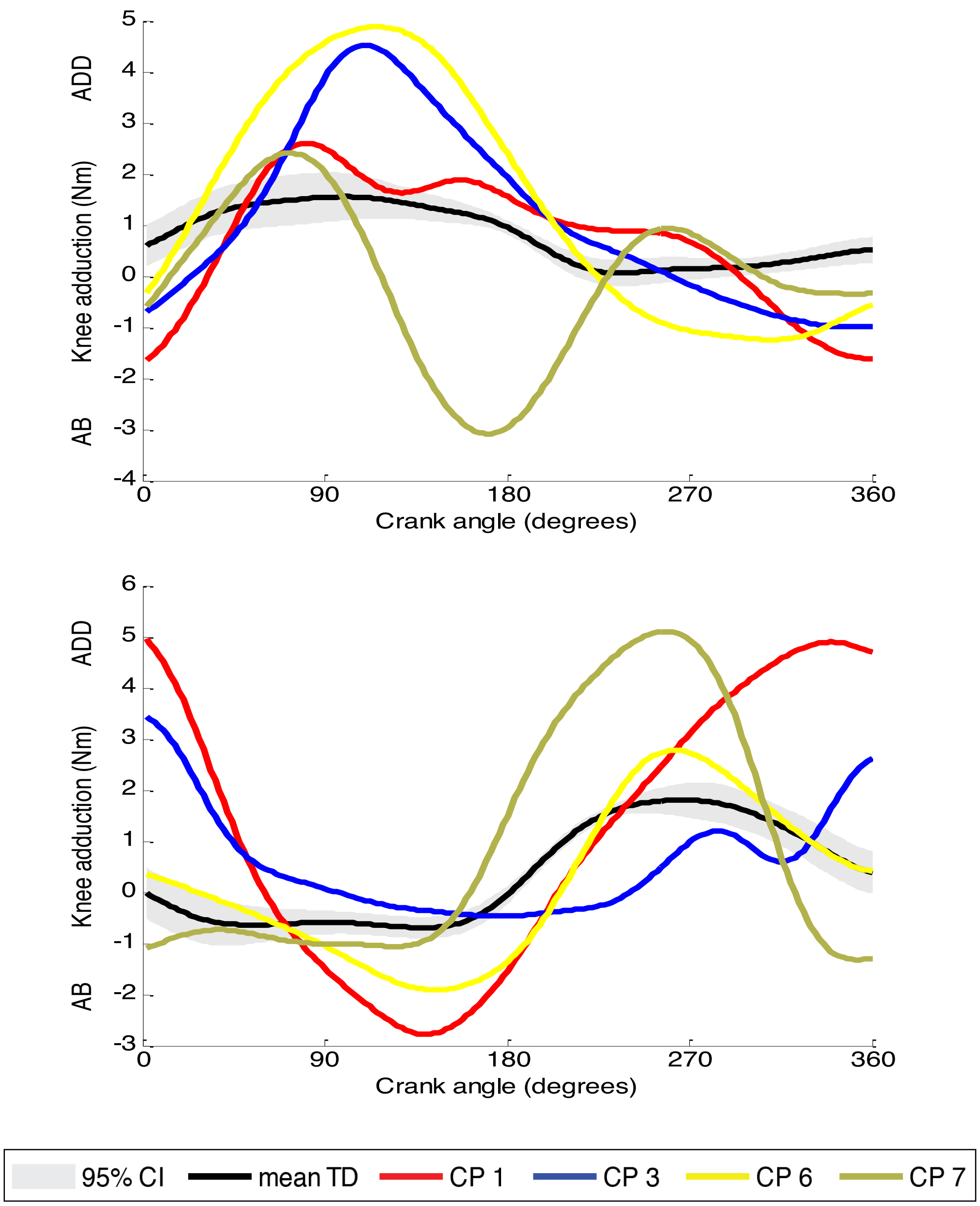


Figure A97: Knee adduction torques at 50 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION TORQUES

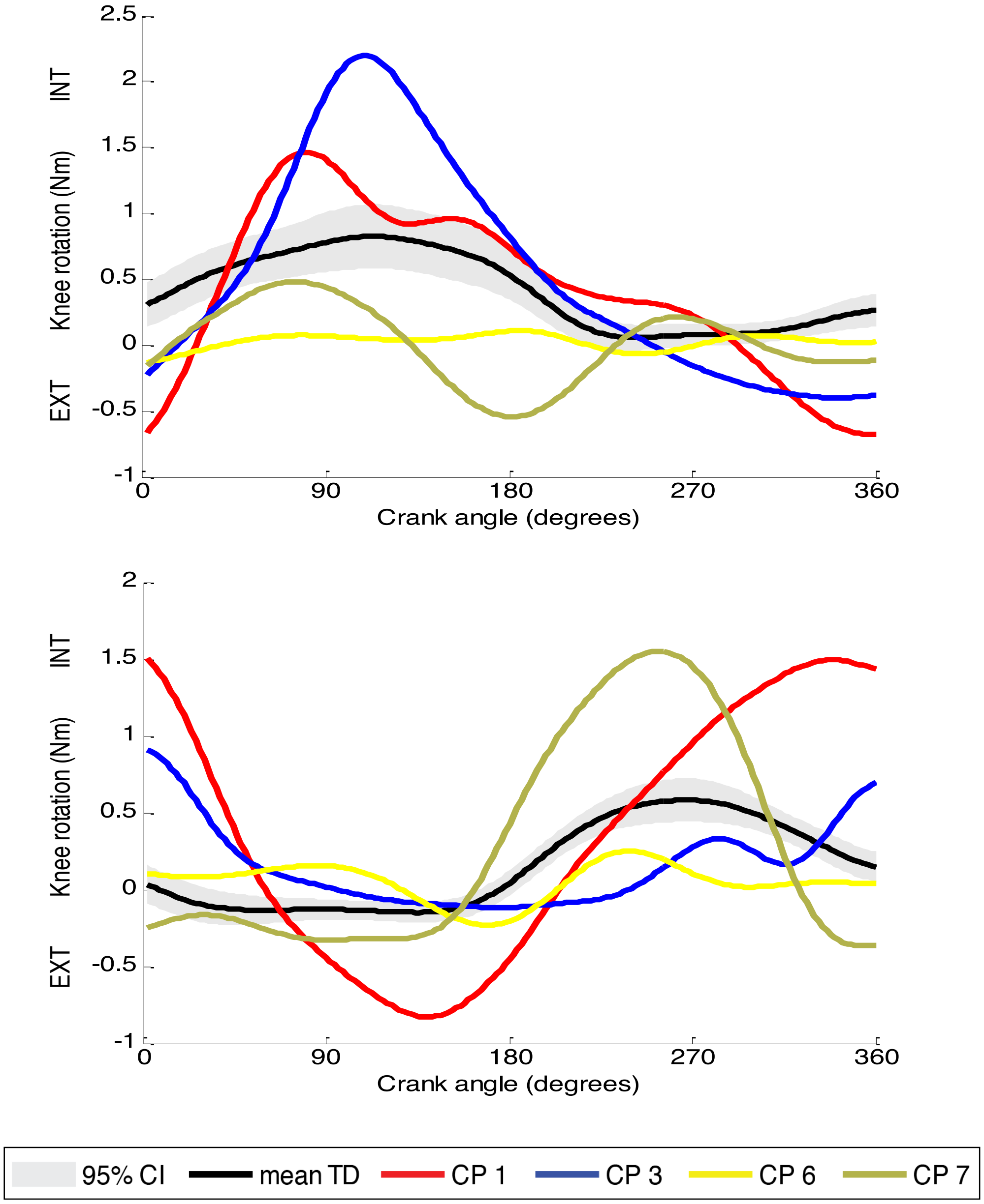


Figure A98: Knee rotation torques at 50 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION TORQUES

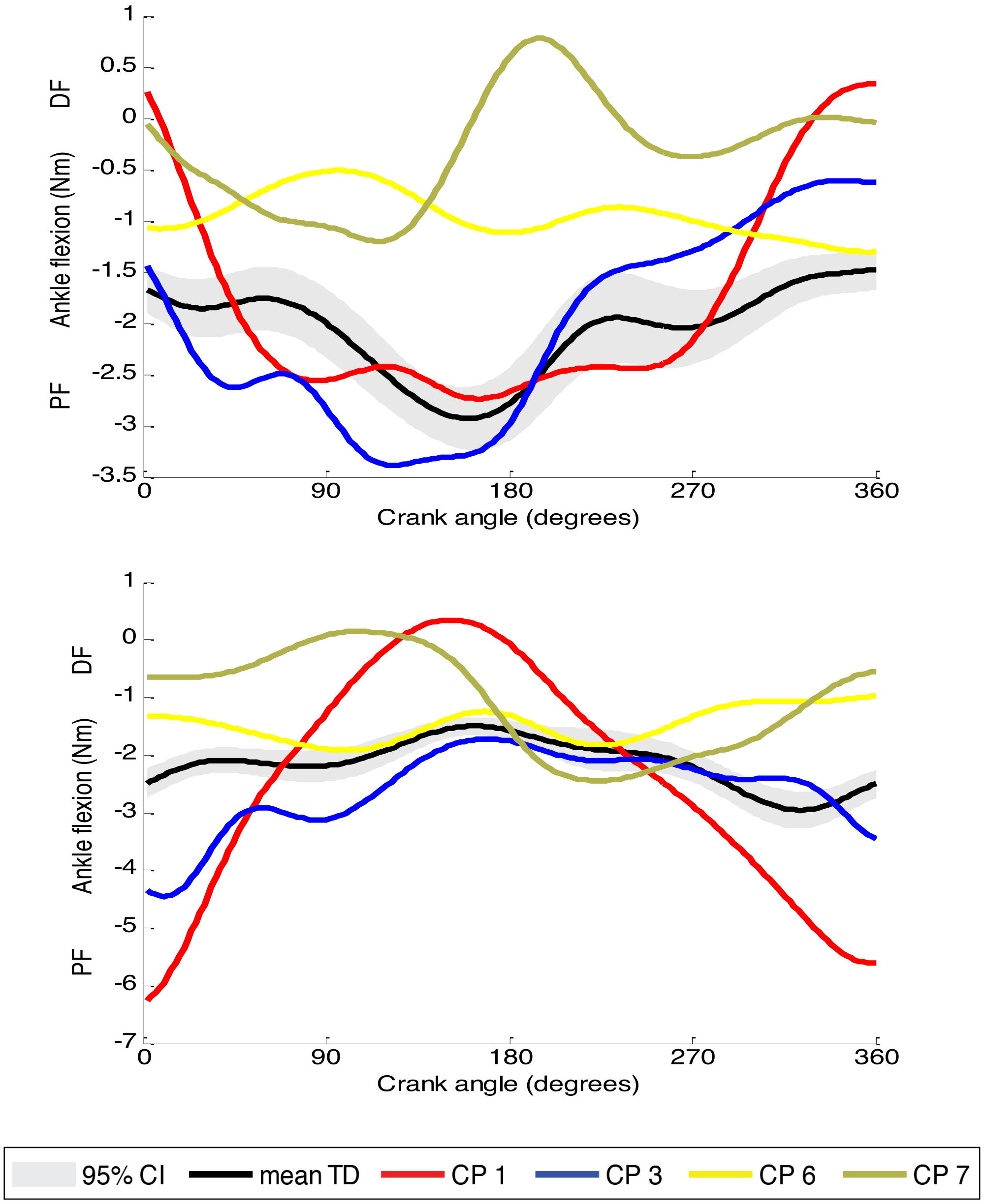


Figure A99: Ankle flexion torques at 50 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION TORQUES

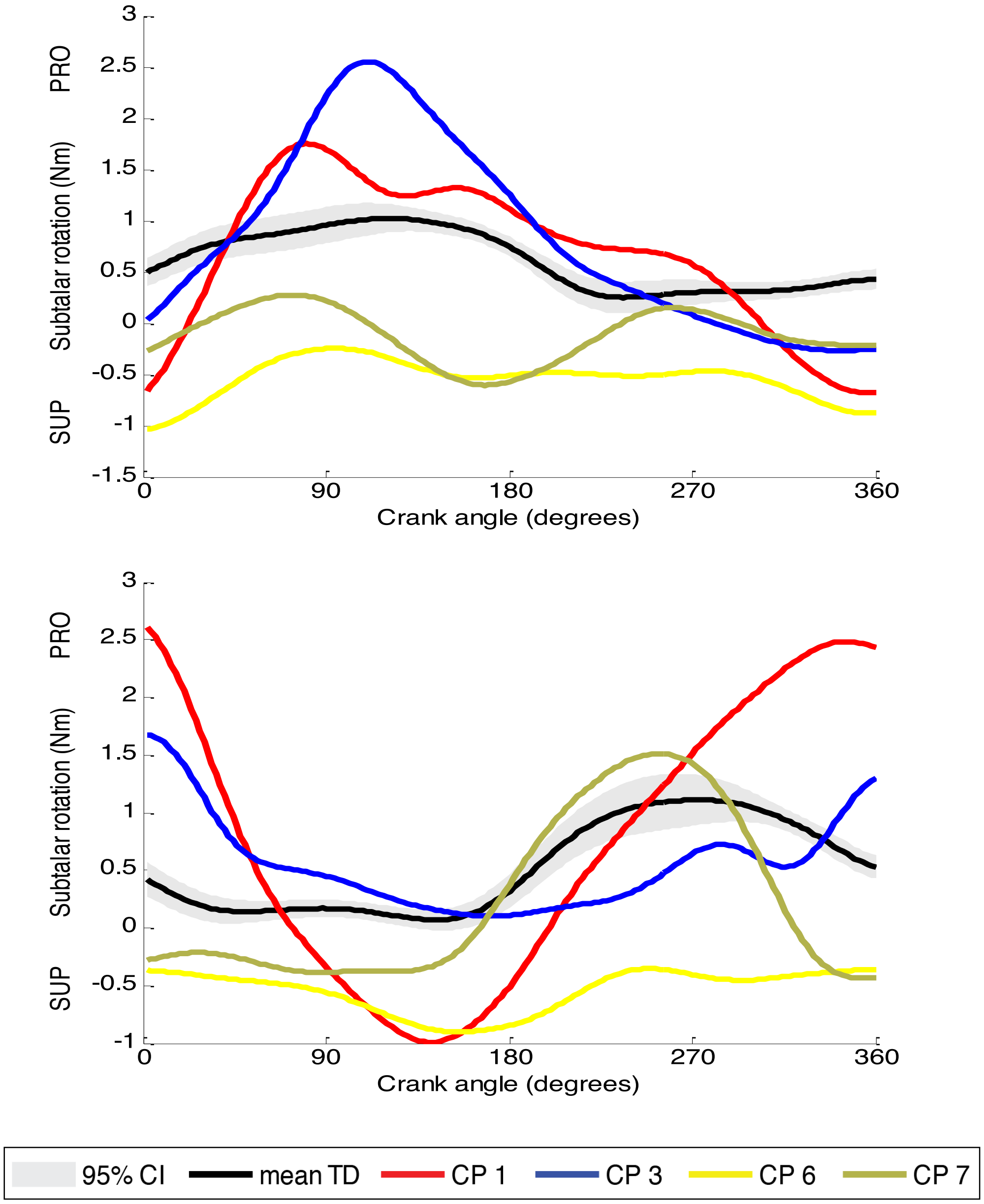


Figure A100: Subtalar rotation torques at 50 RPM produced by right (top) and left (bottom) leg.

APPENDIX AB: JOINT TORQUES AT 60 RPM IN STUDY 2

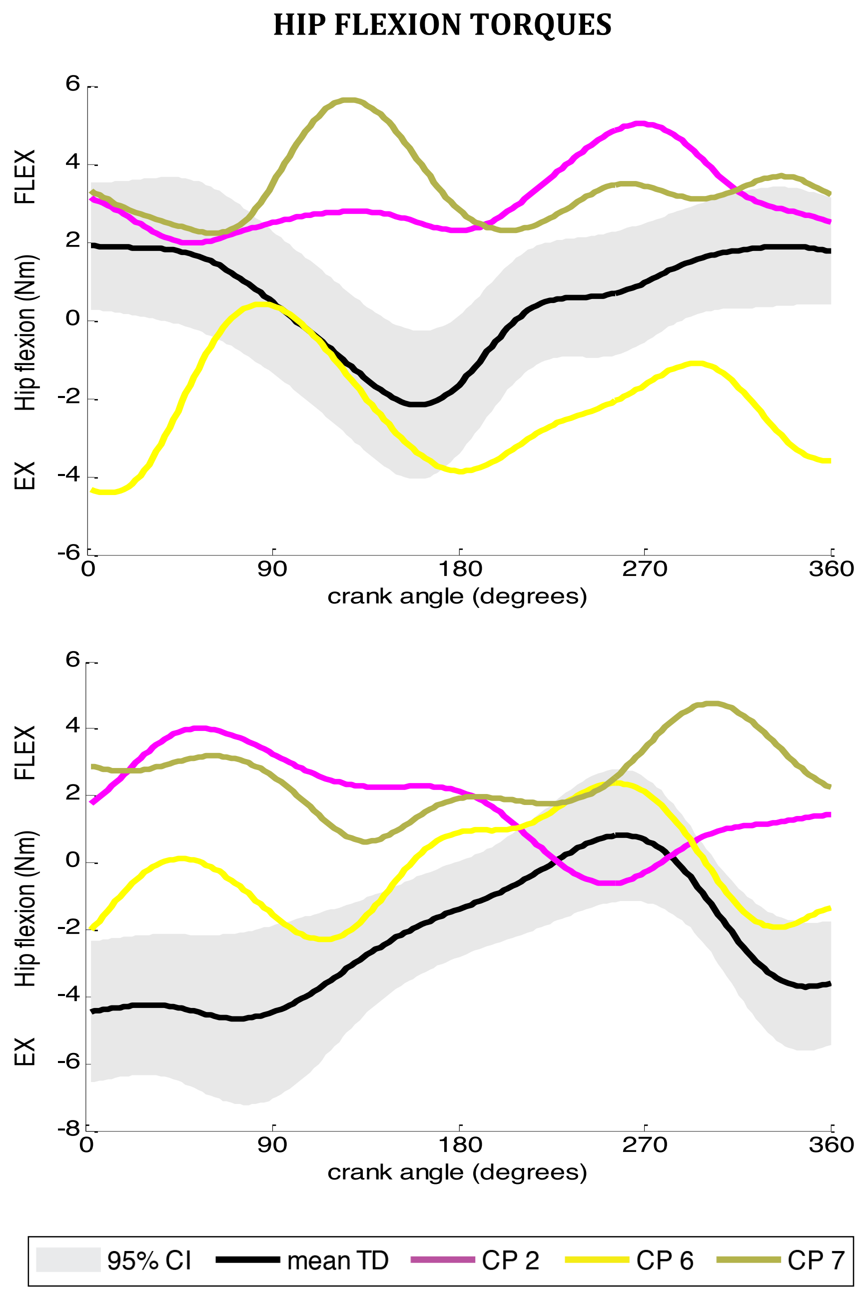


Figure A101: Hip flexion torques at 60 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION TORQUES

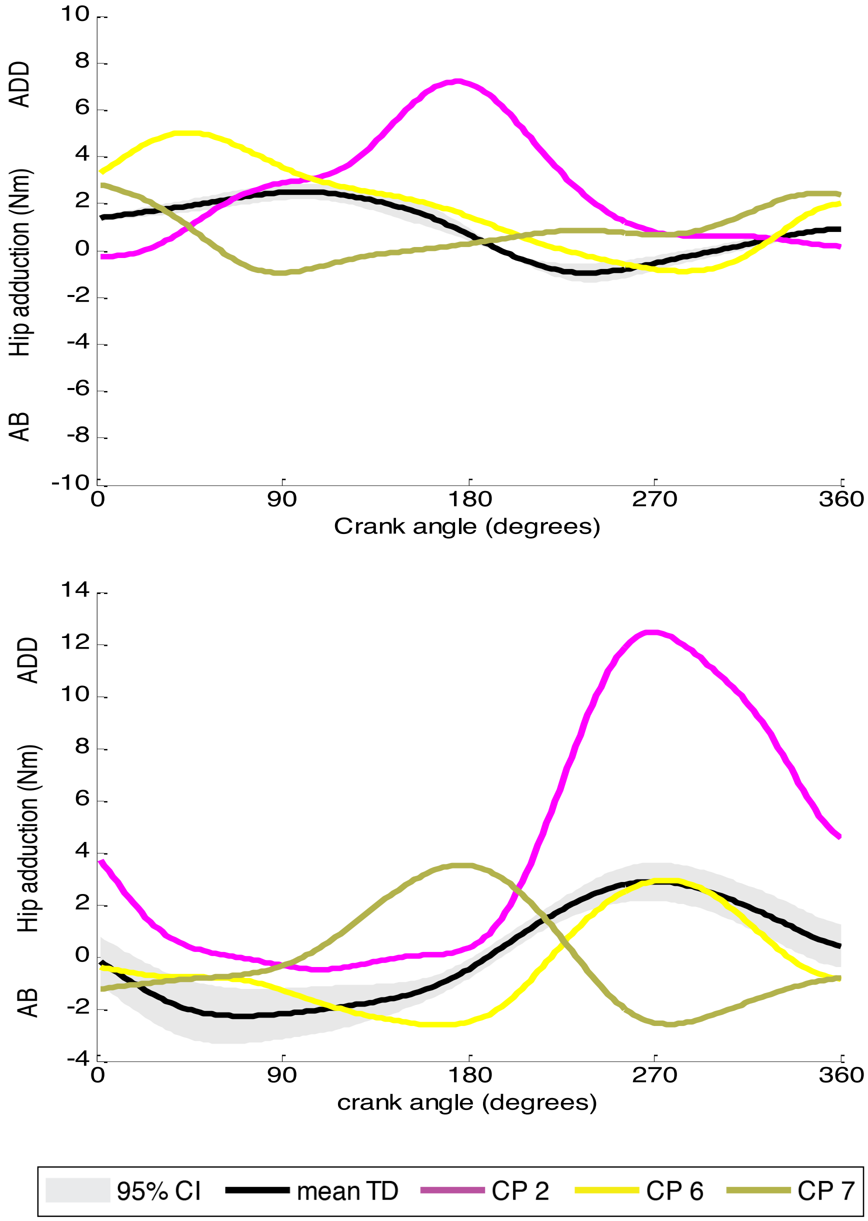


Figure A102: Hip adduction torques at 60 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION TORQUES

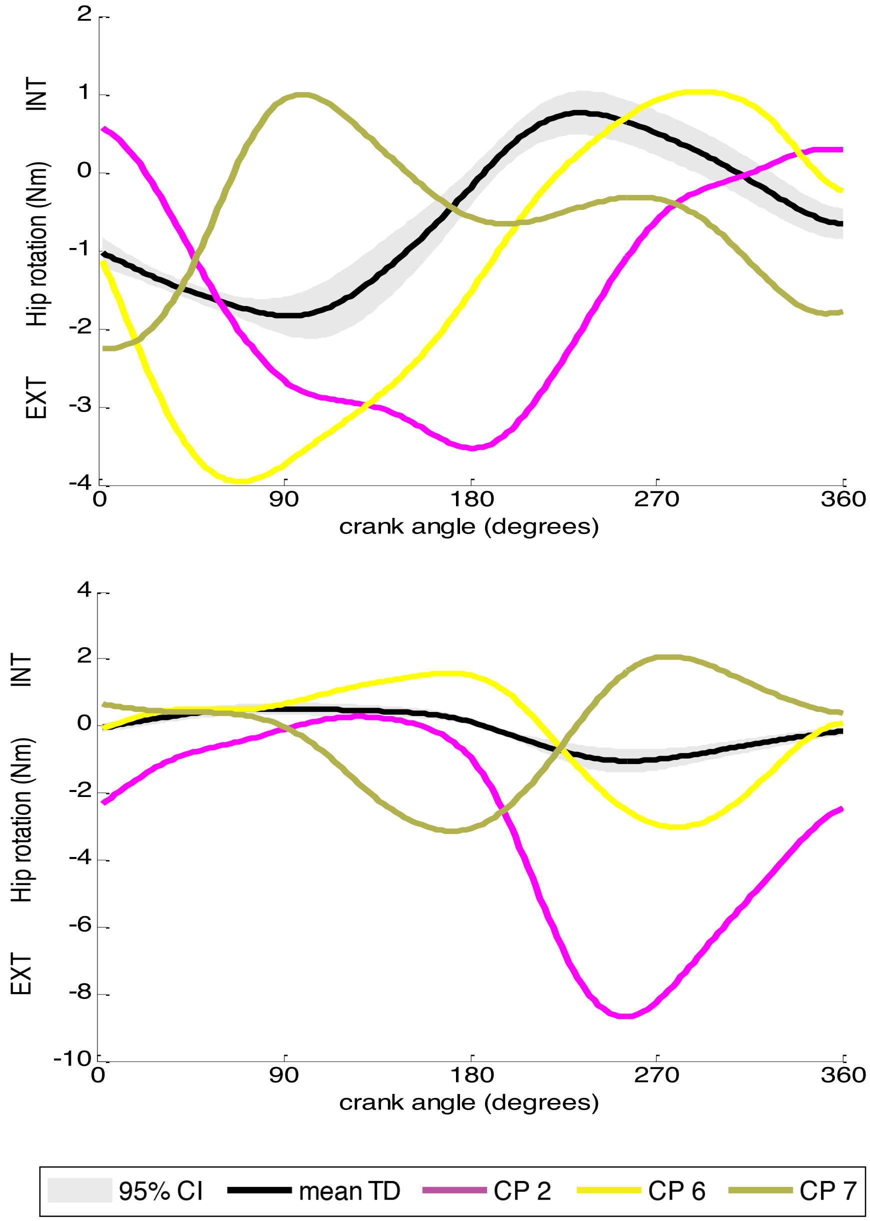


Figure A103: Hip rotation torques at 60 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION TORQUES

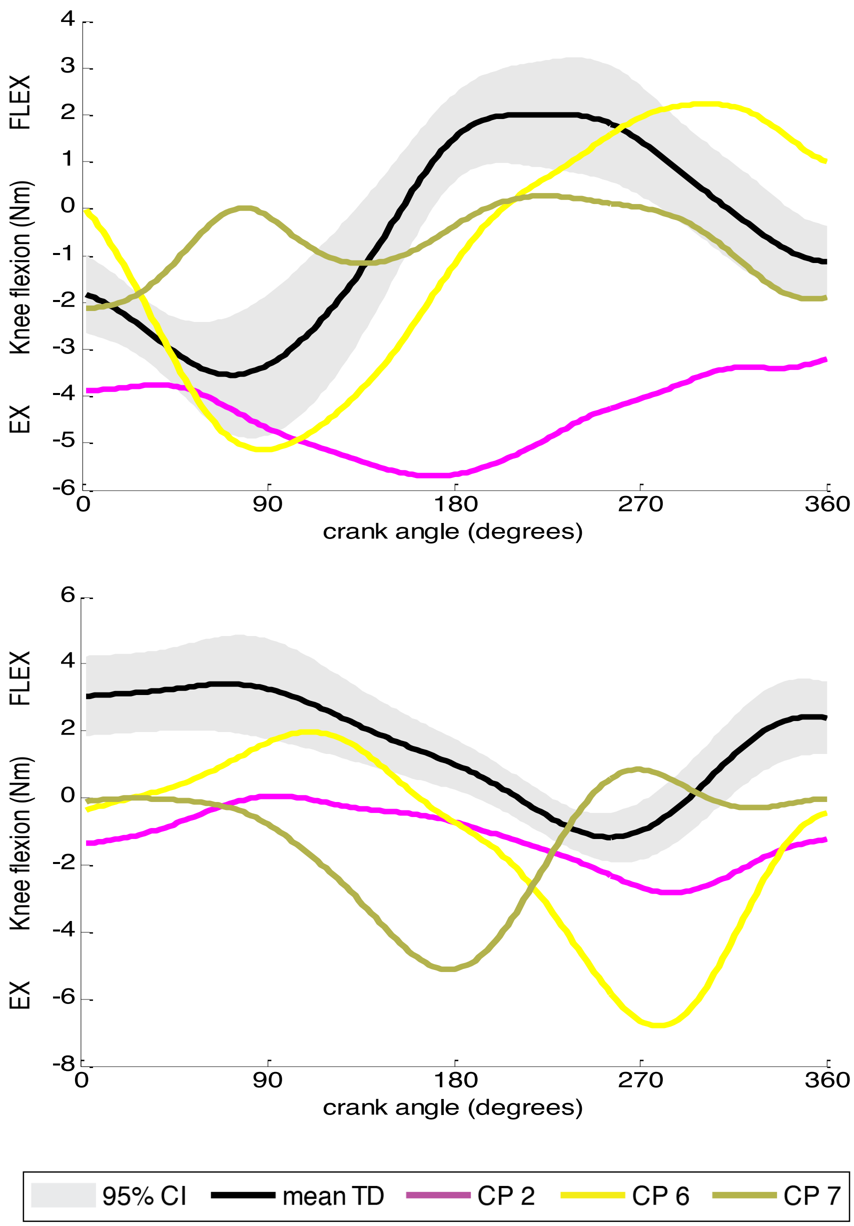


Figure A104: Knee flexion torques at 60 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION TORQUES

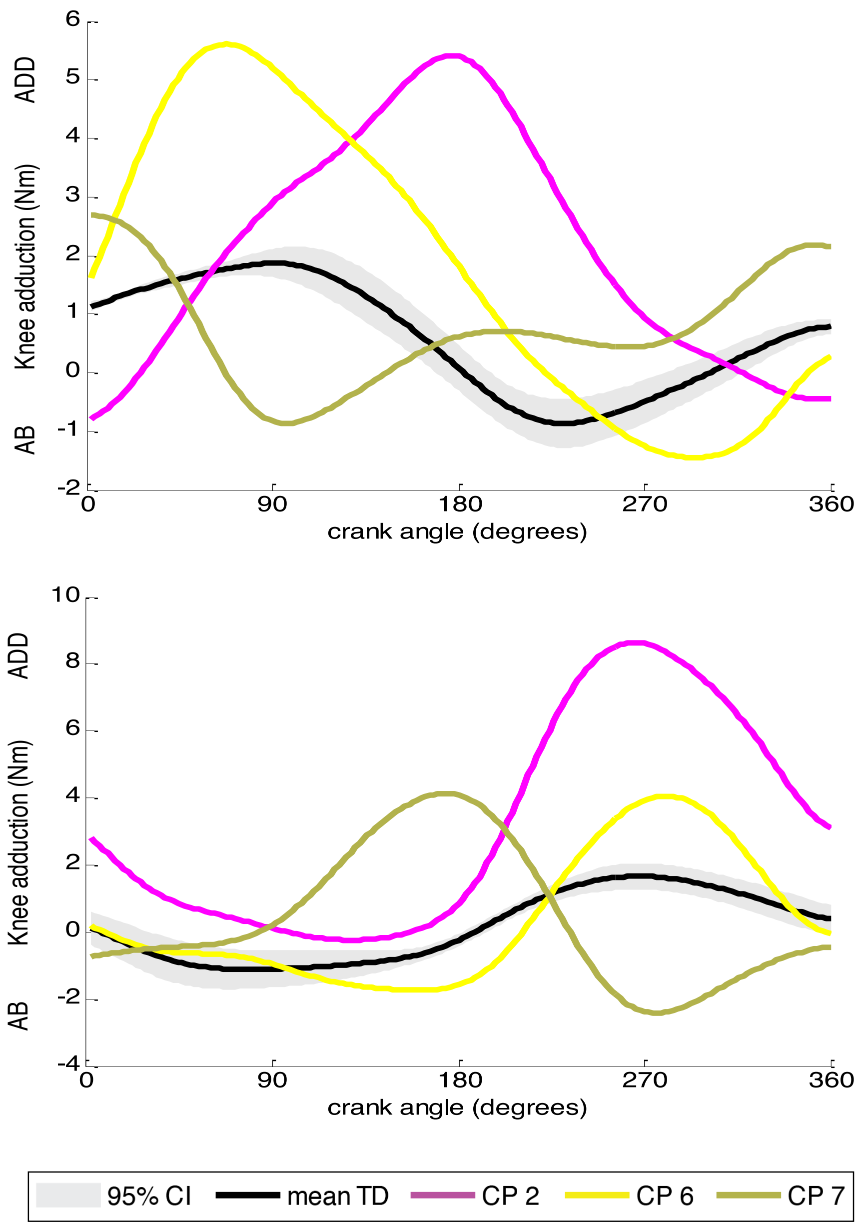


Figure A105: Knee adduction torques at 60 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION TORQUES

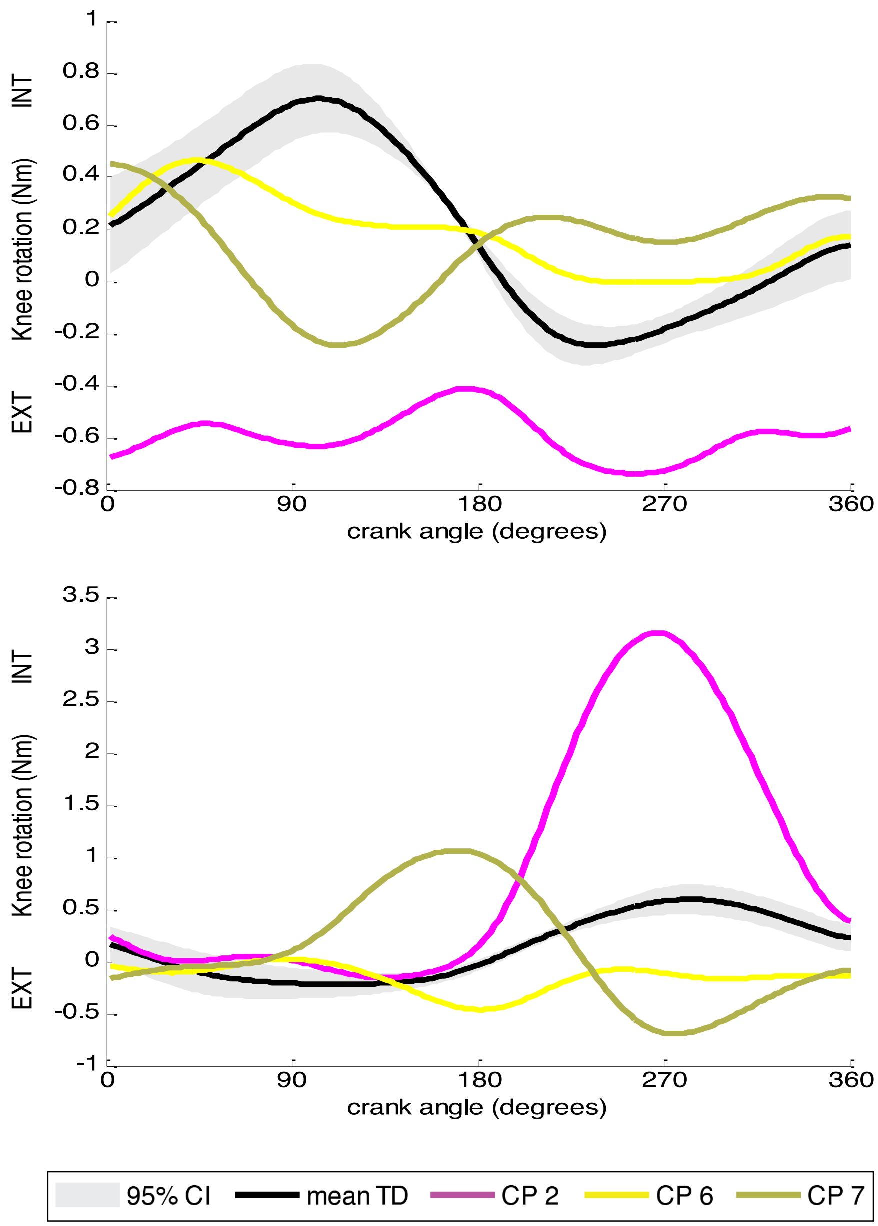


Figure A106: Knee rotation torques at 60 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION TORQUES

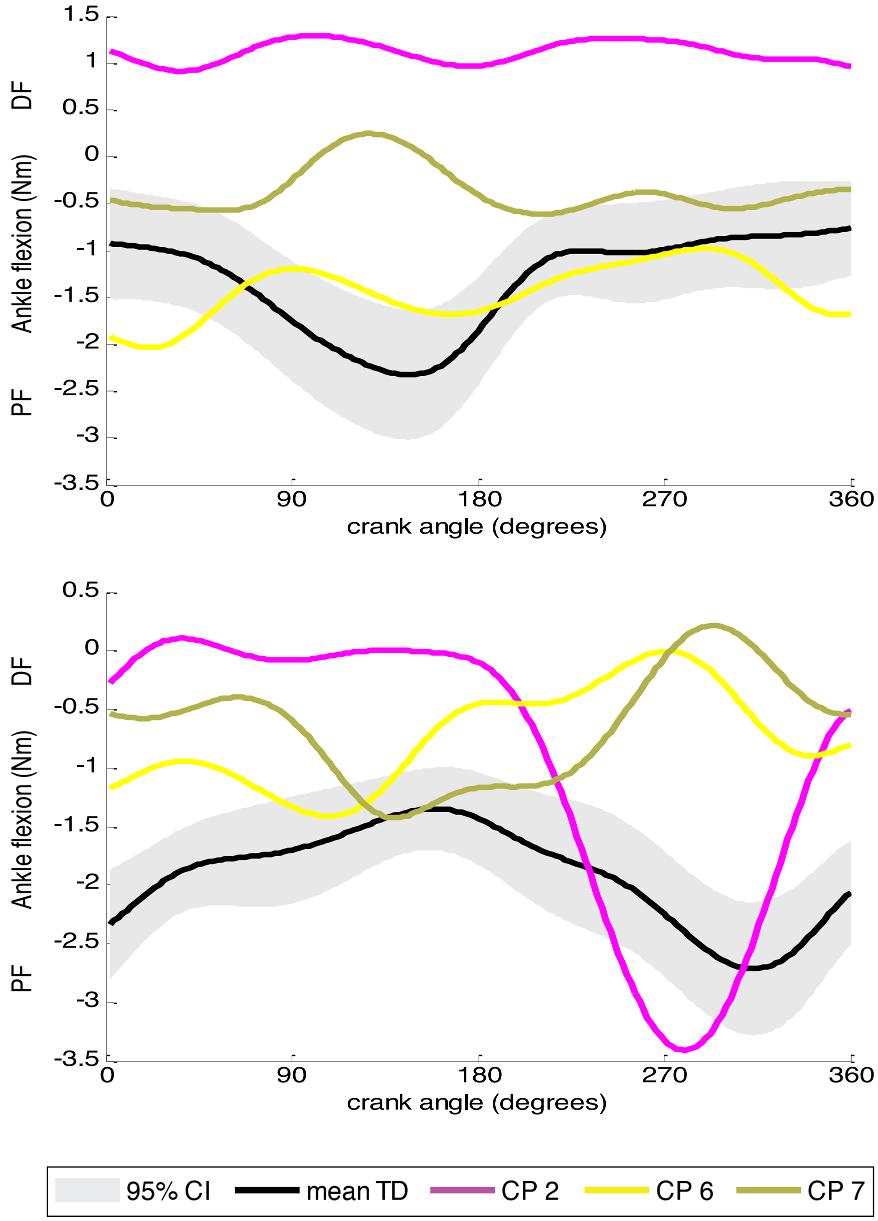


Figure A107: Ankle flexion torques at 60 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION TORQUES

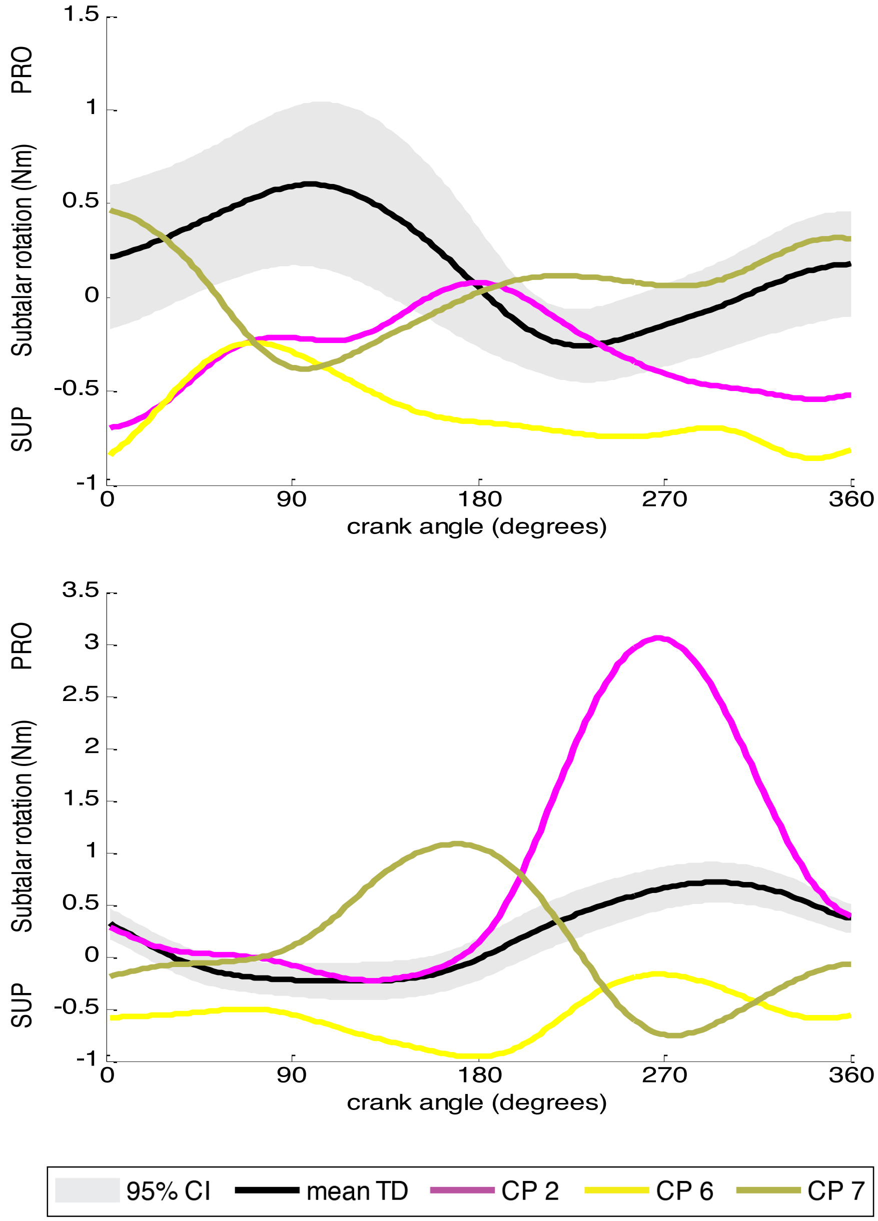


Figure A108: Subtalar rotation torques at 60 RPM produced by right (top) and left (bottom) leg.

APPENDIX AC: JOINT TORQUES AT 70 RPM IN STUDY 2

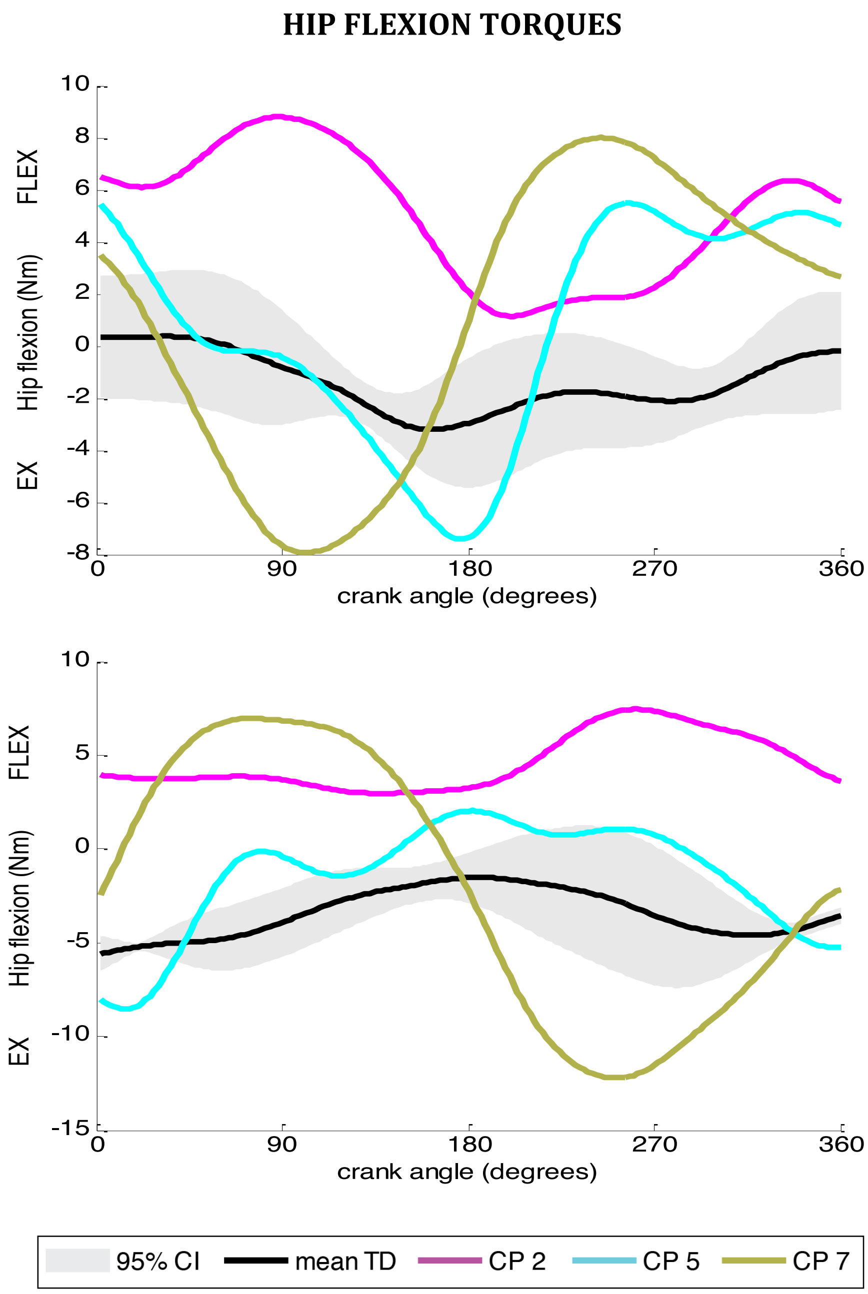


Figure A109: Hip flexion torques at 70 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION TORQUES

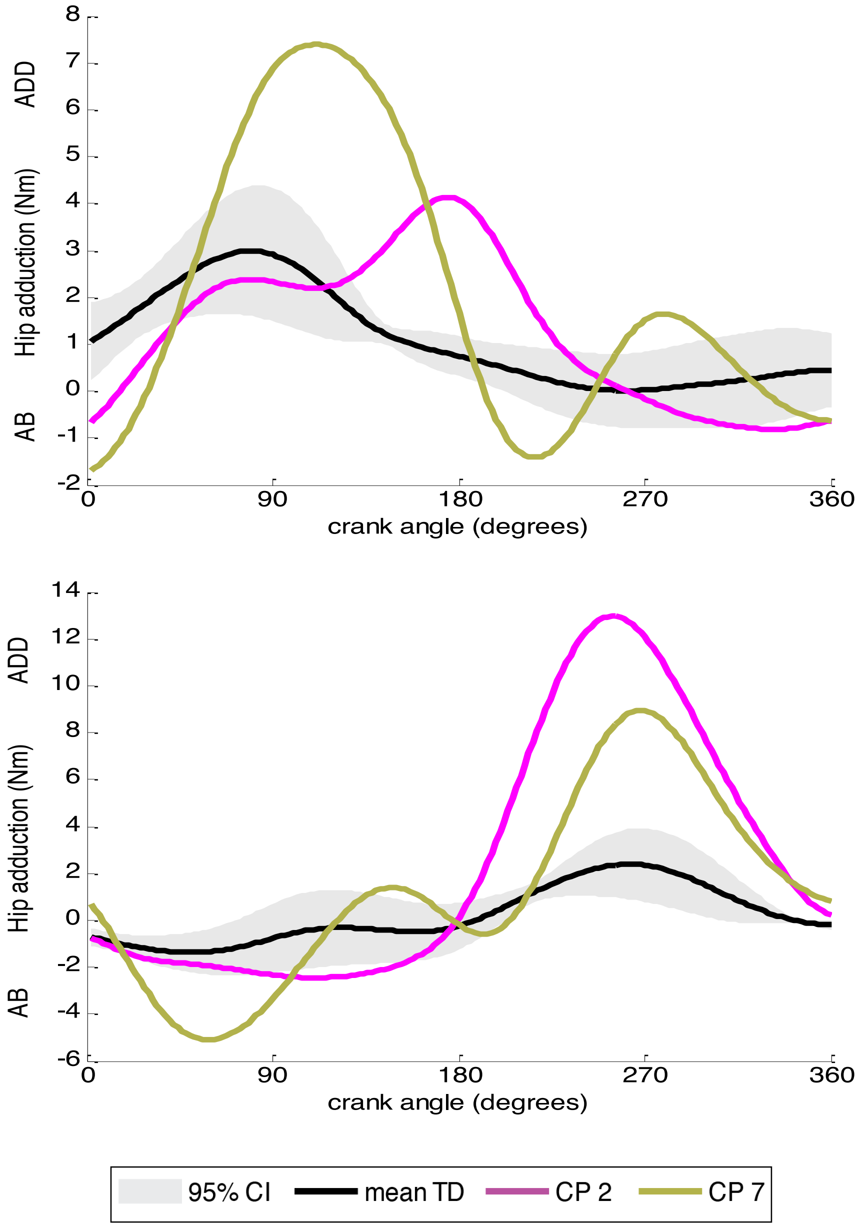


Figure A110: Hip adduction torques at 70 RPM produced by right (top) and left (bottom) leg.

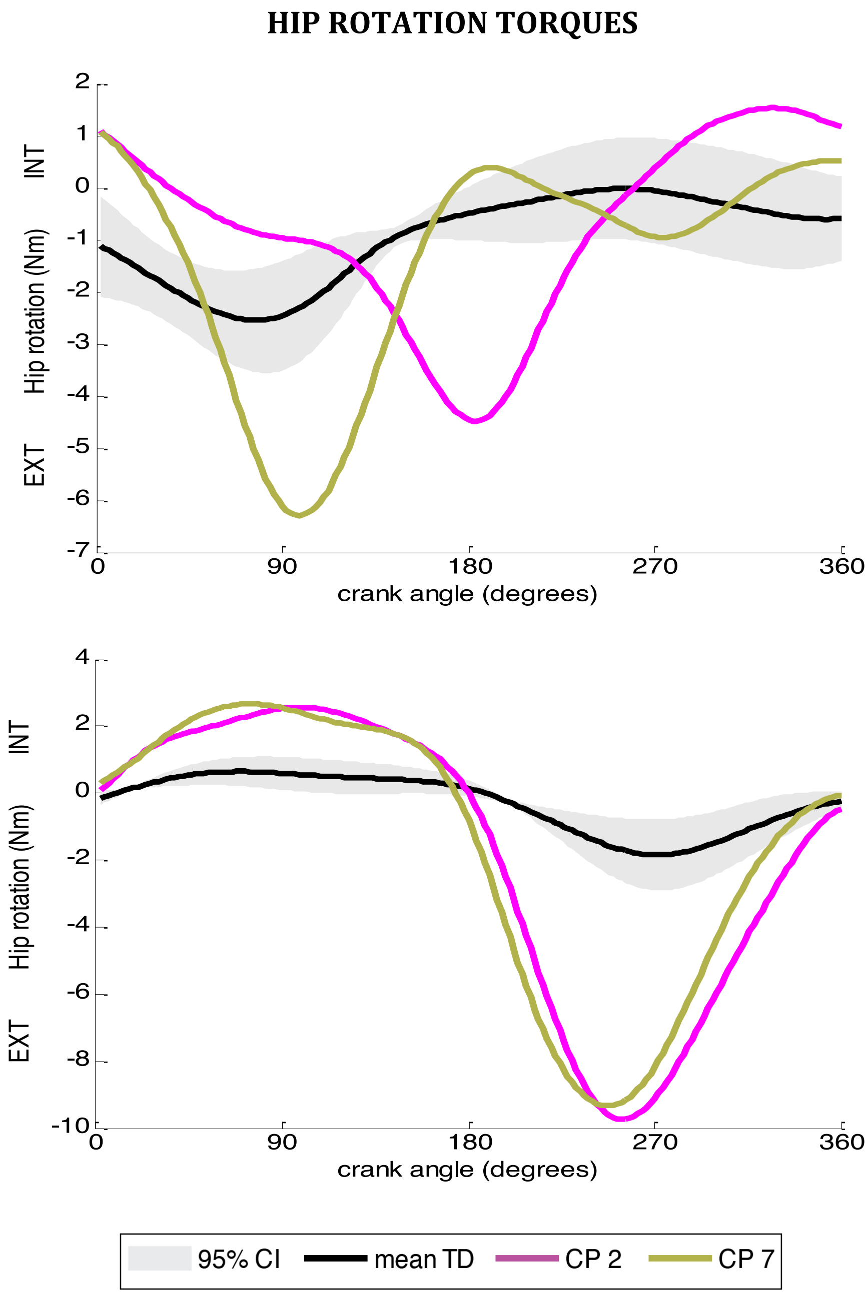


Figure A111: Hip rotation torques at 70 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION TORQUES

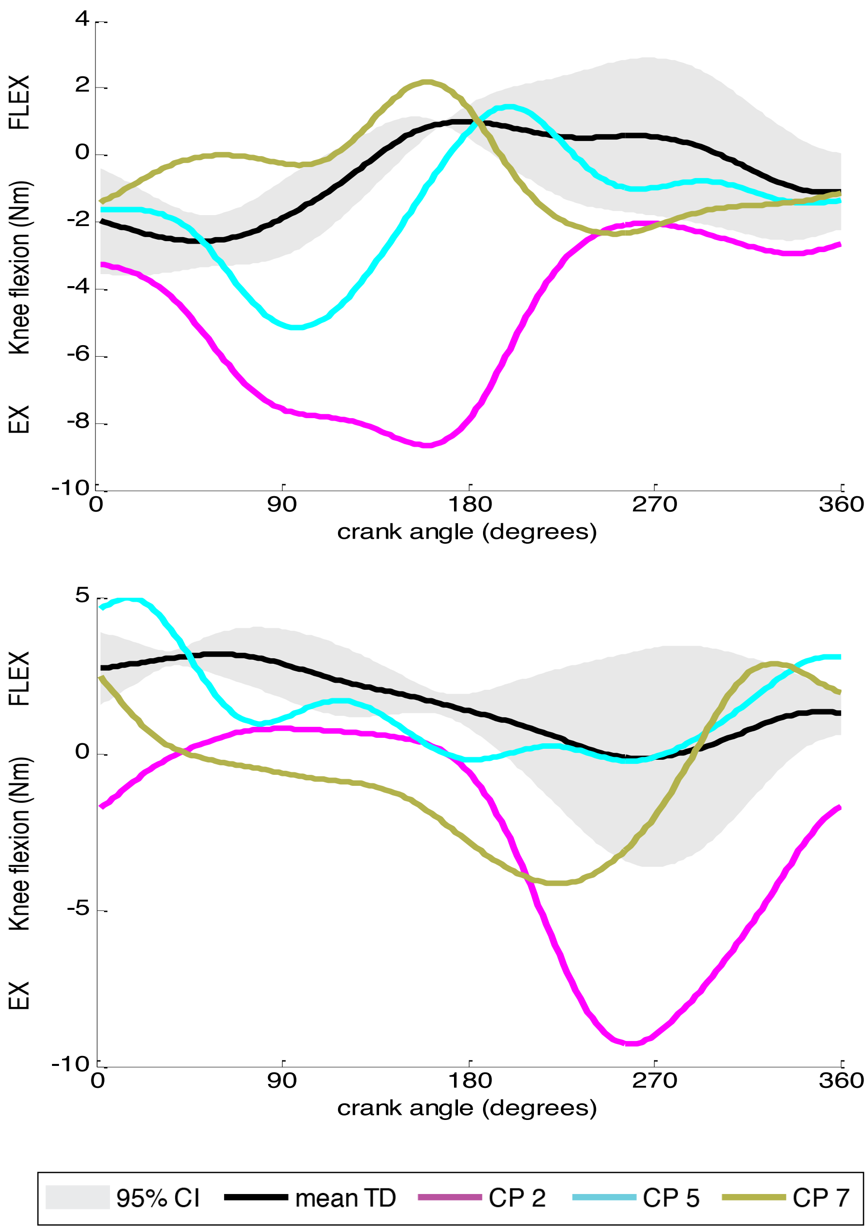


Figure A112: Knee flexion torques at 70 RPM produced by right (top) and left (bottom) leg.

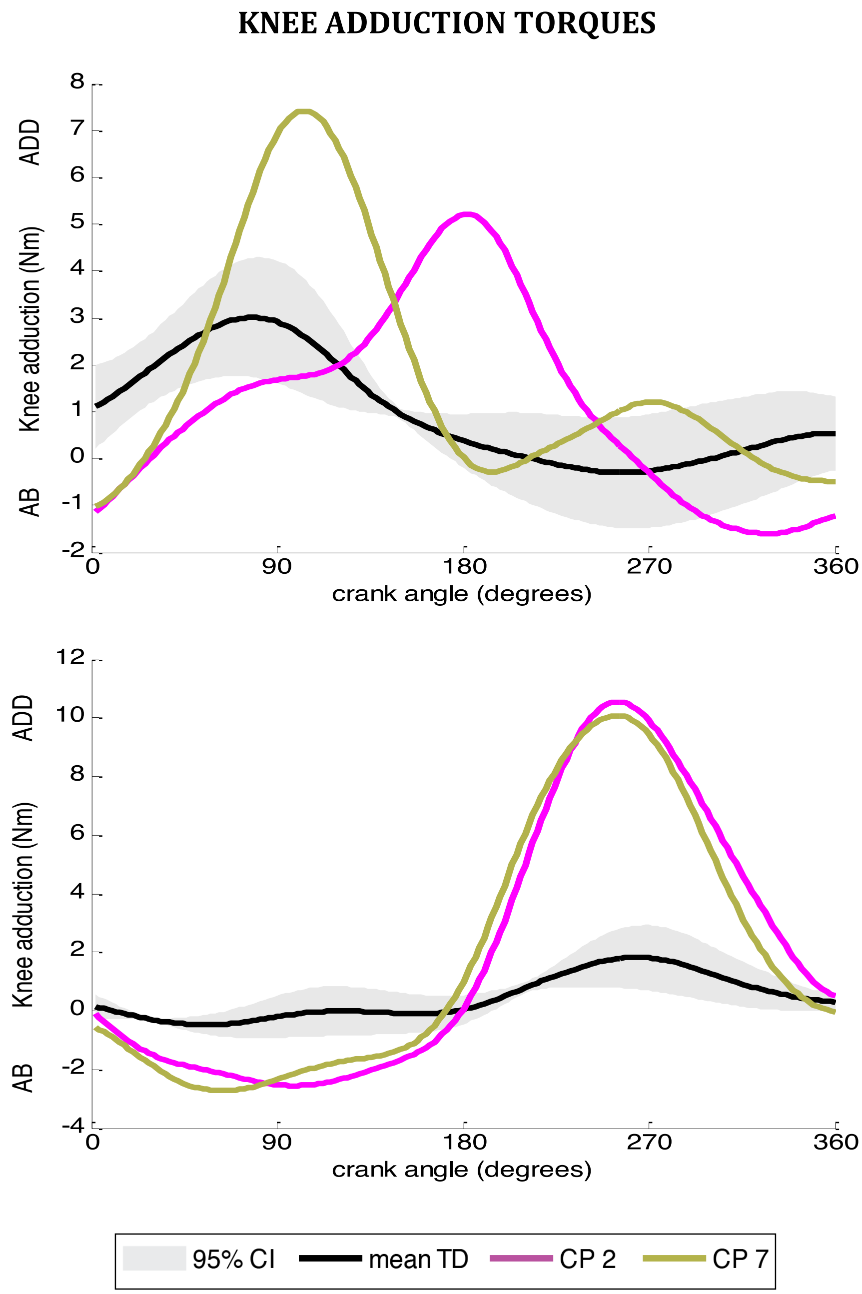


Figure A113: Knee adduction torques at 70 RPM produced by right (top) and left (bottom) leg.

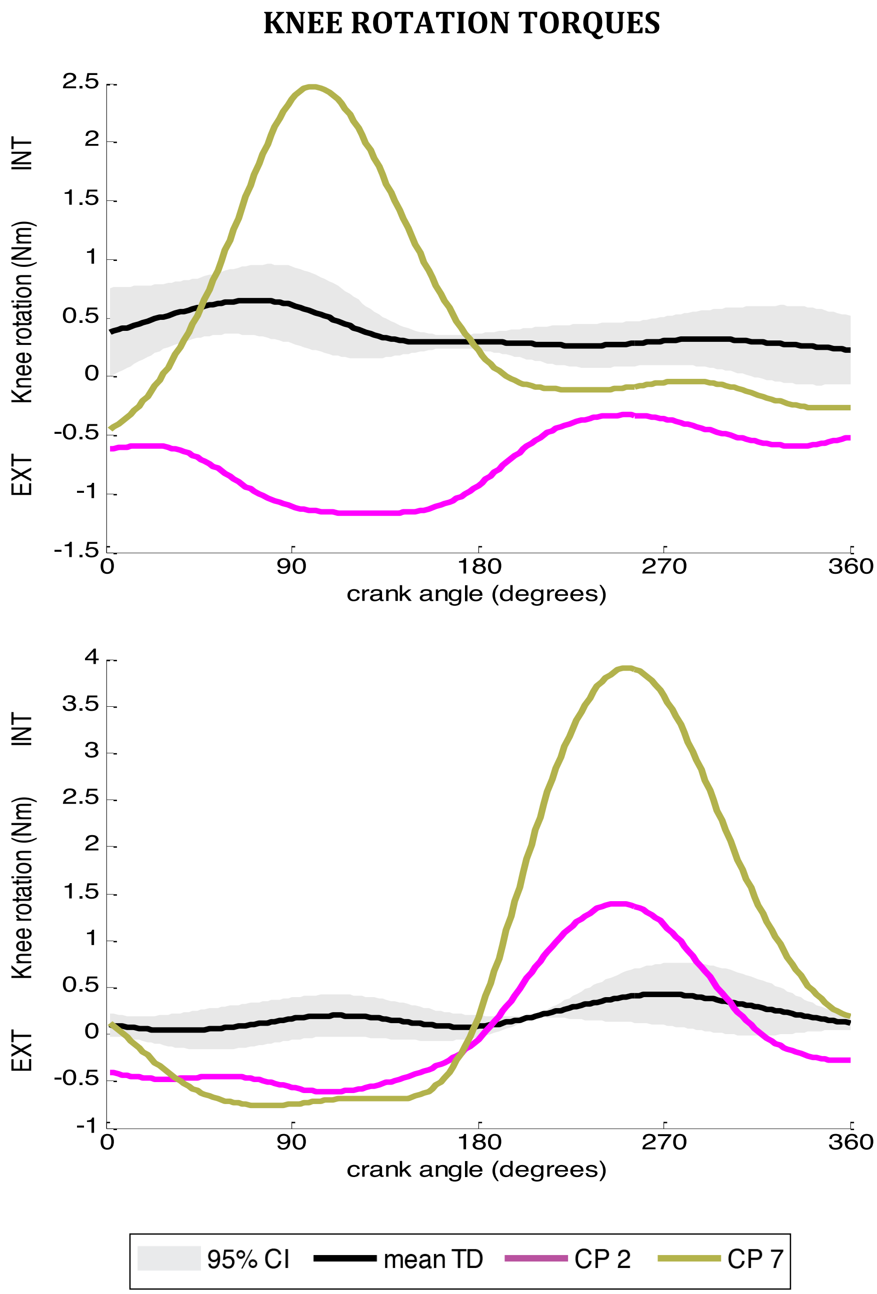


Figure A114: Knee rotation torques at 70 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION TORQUES

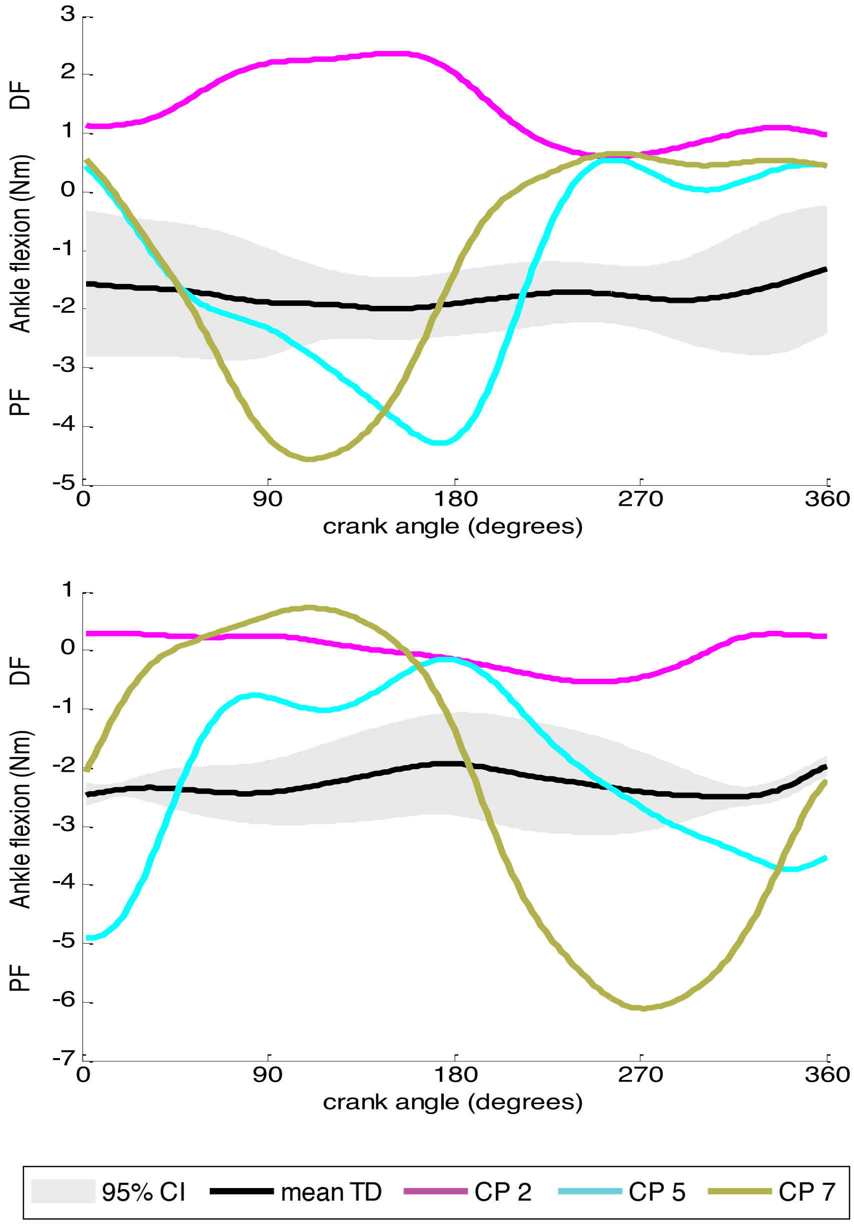


Figure A115: Ankle flexion torques at 70 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION TORQUES

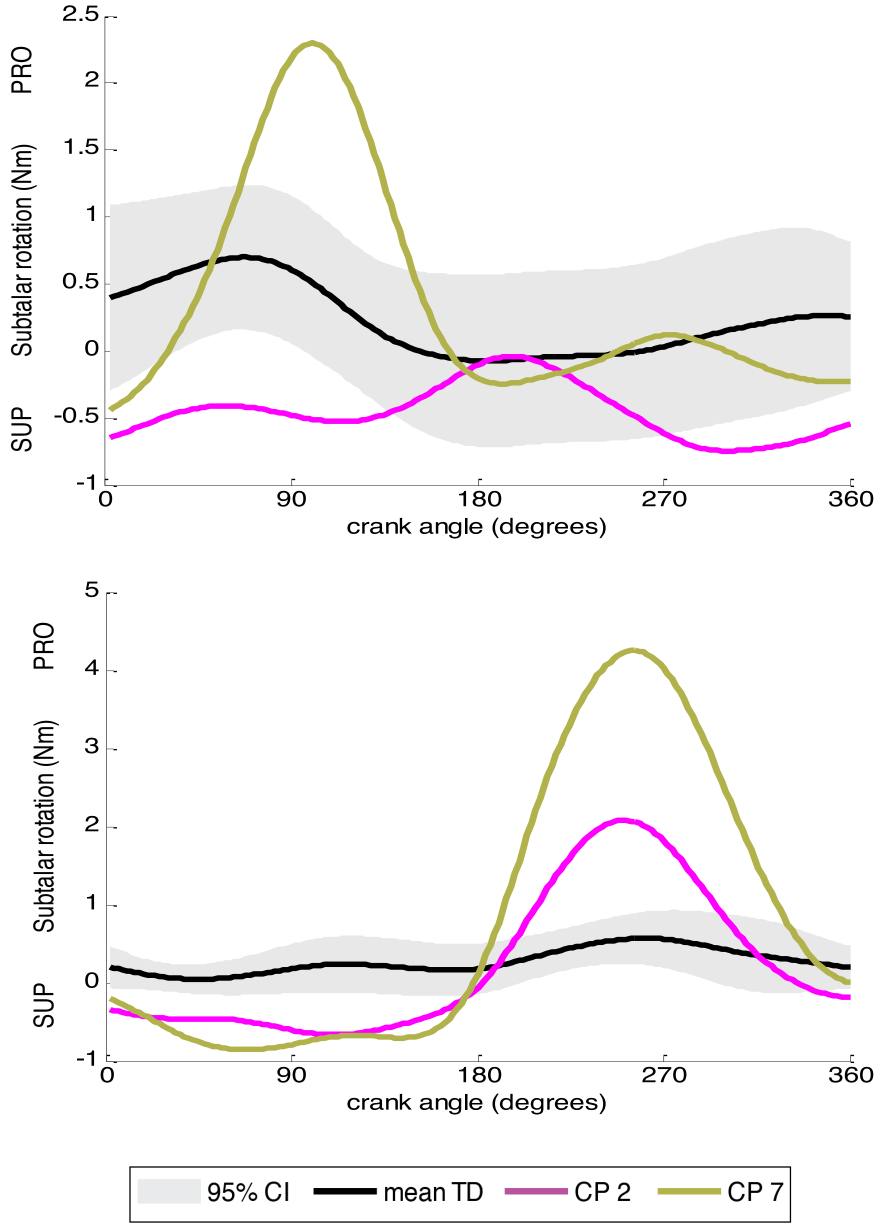


Figure A116: Subtalar rotation torques at 70 RPM produced by right (top) and left (bottom) leg.

APPENDIX AD: JOINT TORQUES AT 80 RPM IN STUDY 2

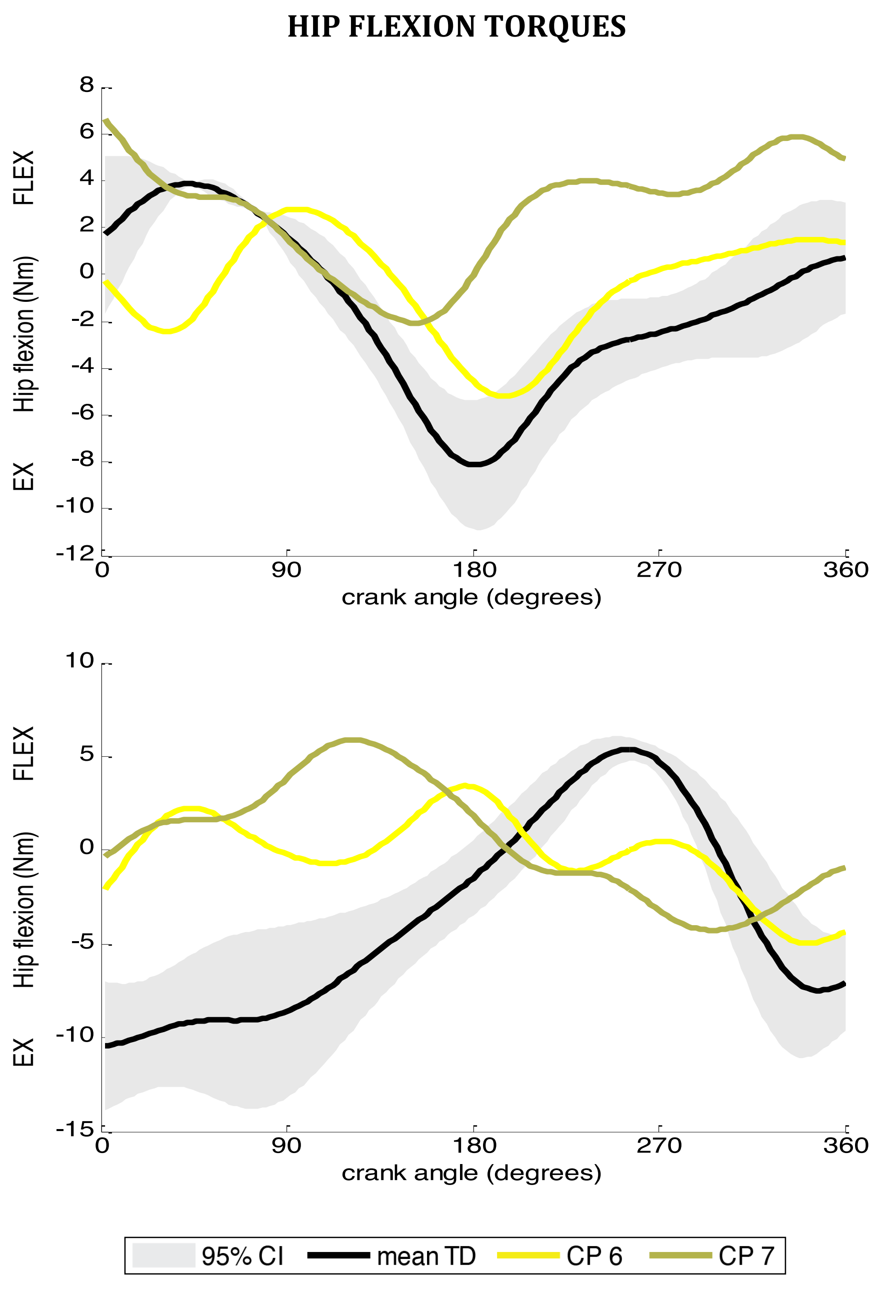


Figure A117: Hip flexion torques at 80 RPM produced by right (top) and left (bottom) leg.

HIP ADDUCTION TORQUES

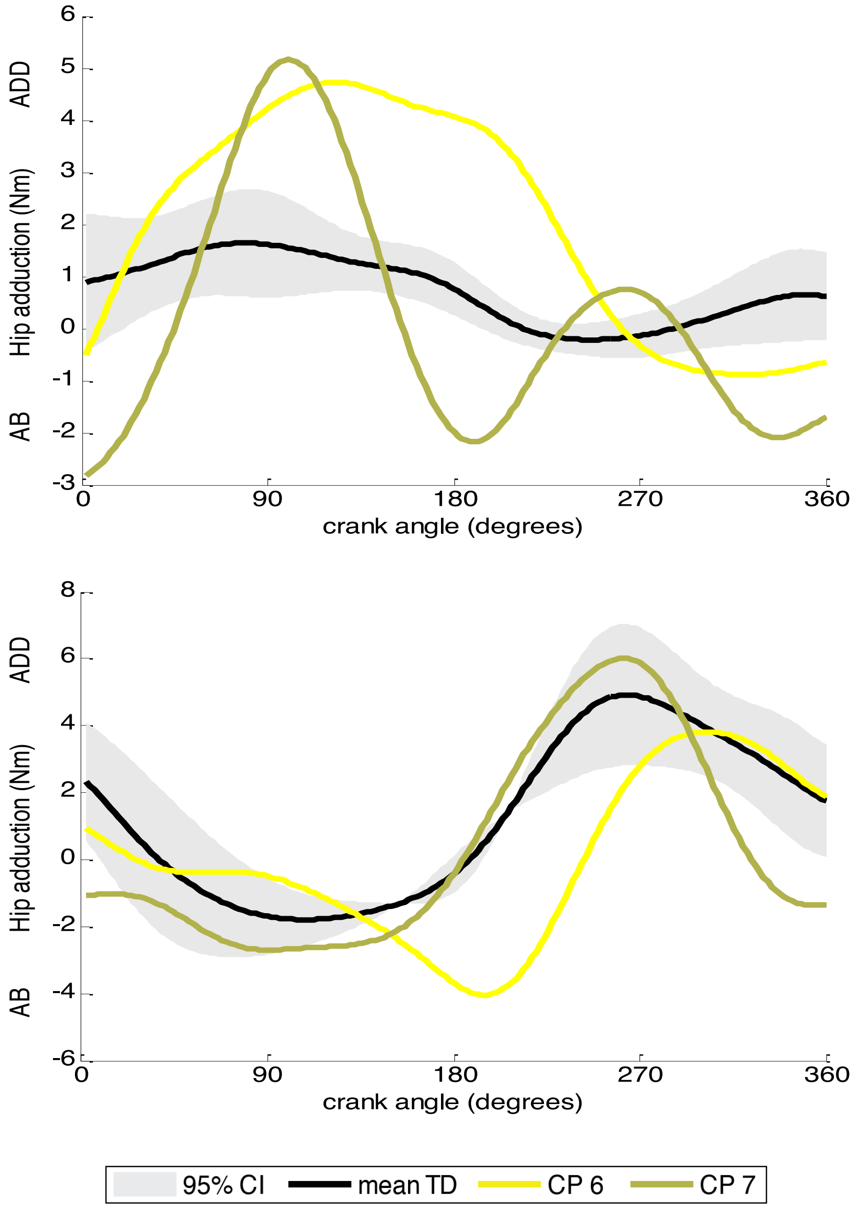


Figure A118: Hip adduction torques at 80 RPM produced by right (top) and left (bottom) leg.

HIP ROTATION TORQUES

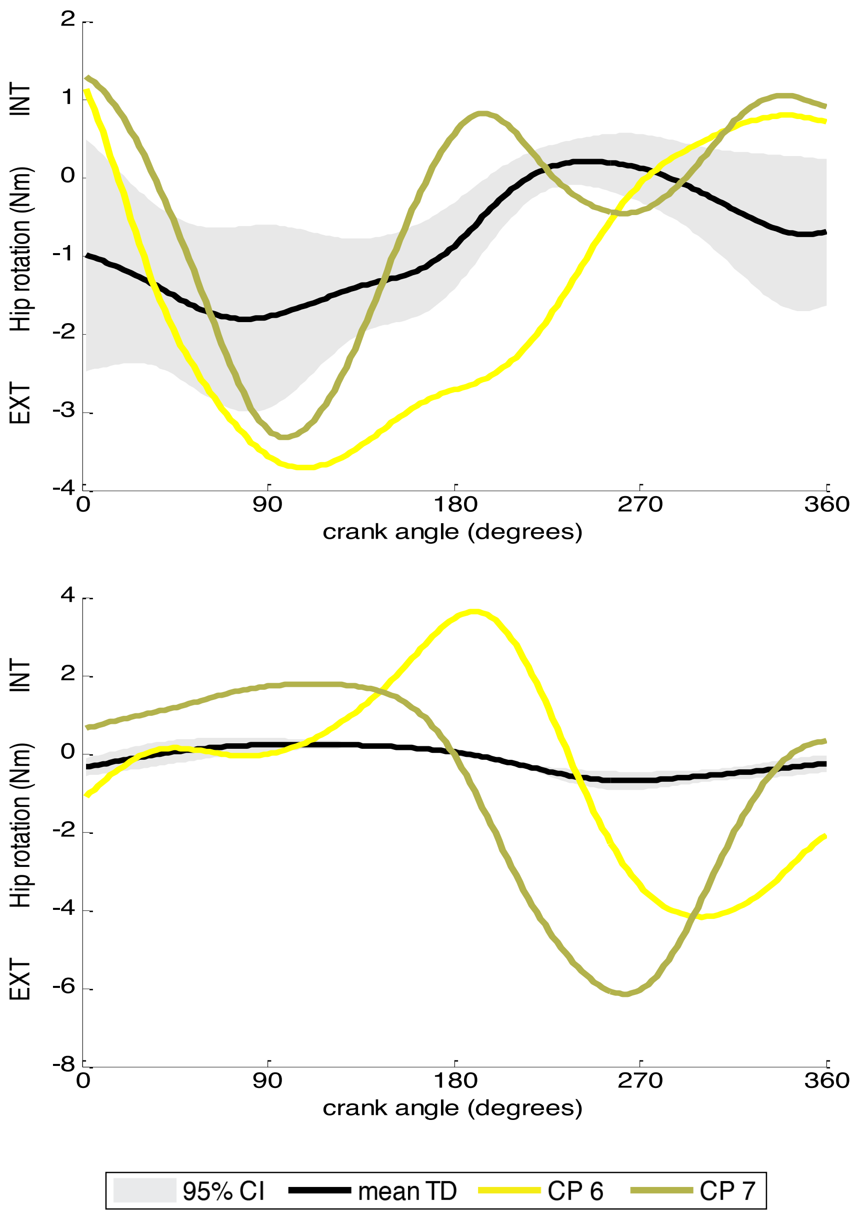


Figure A119: Hip rotation torques at 80 RPM produced by right (top) and left (bottom) leg.

KNEE FLEXION TORQUES

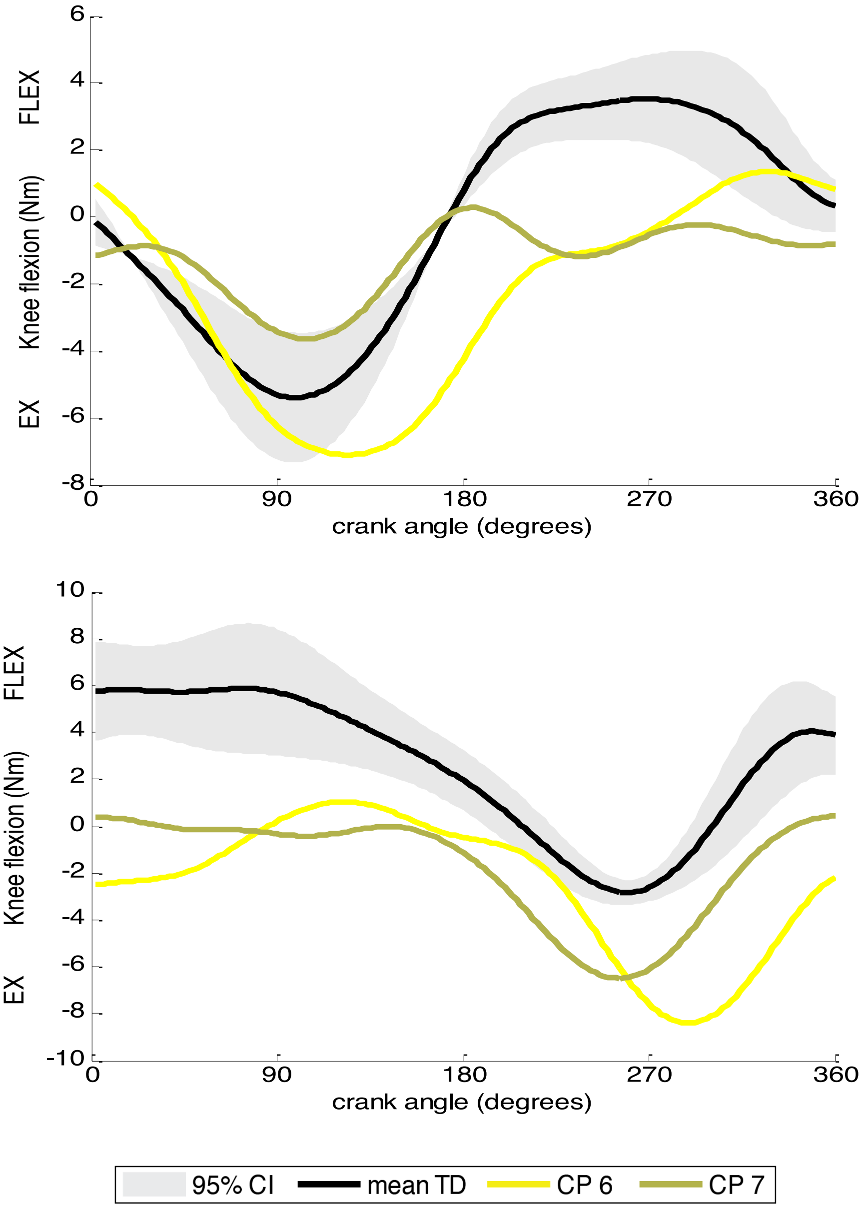


Figure A120: Knee flexion torques at 80 RPM produced by right (top) and left (bottom) leg.

KNEE ADDUCTION TORQUES

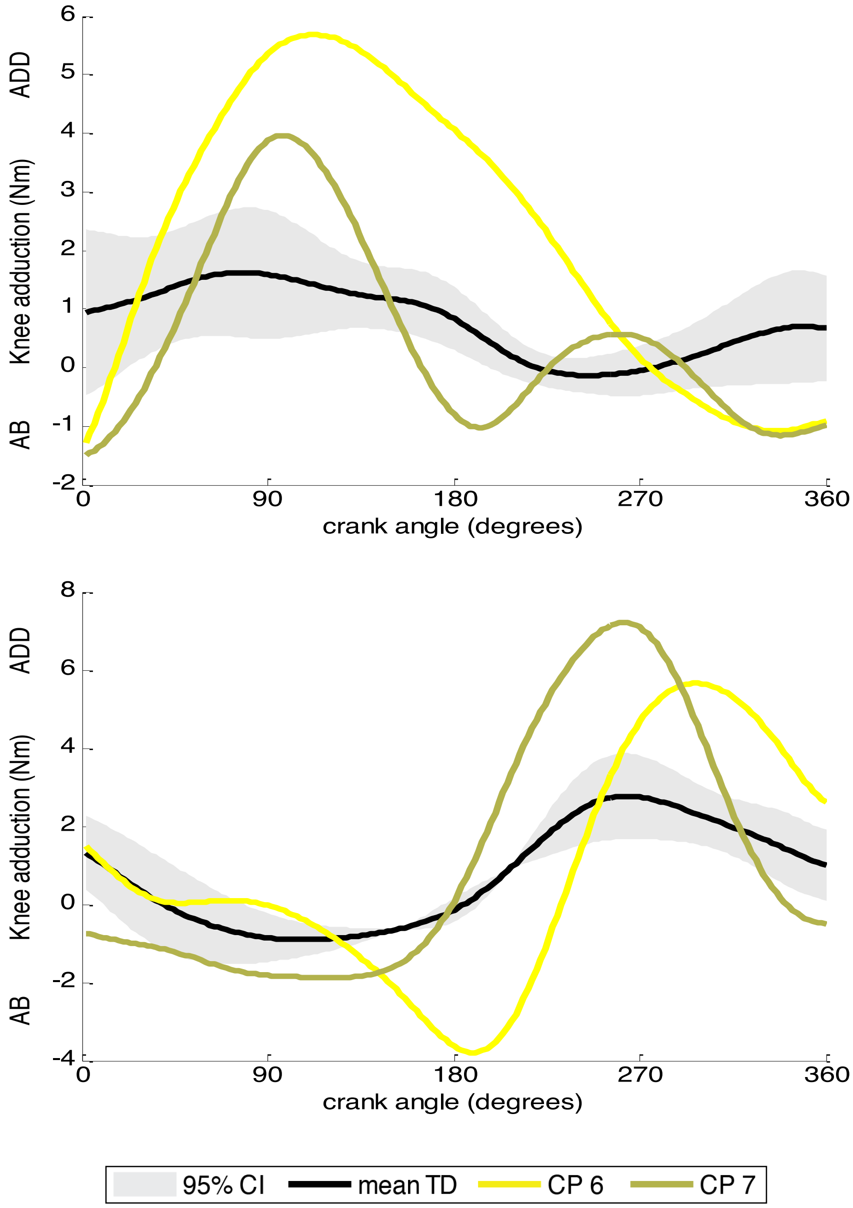


Figure A121: Knee adduction torques at 80 RPM produced by right (top) and left (bottom) leg.

KNEE ROTATION TORQUES

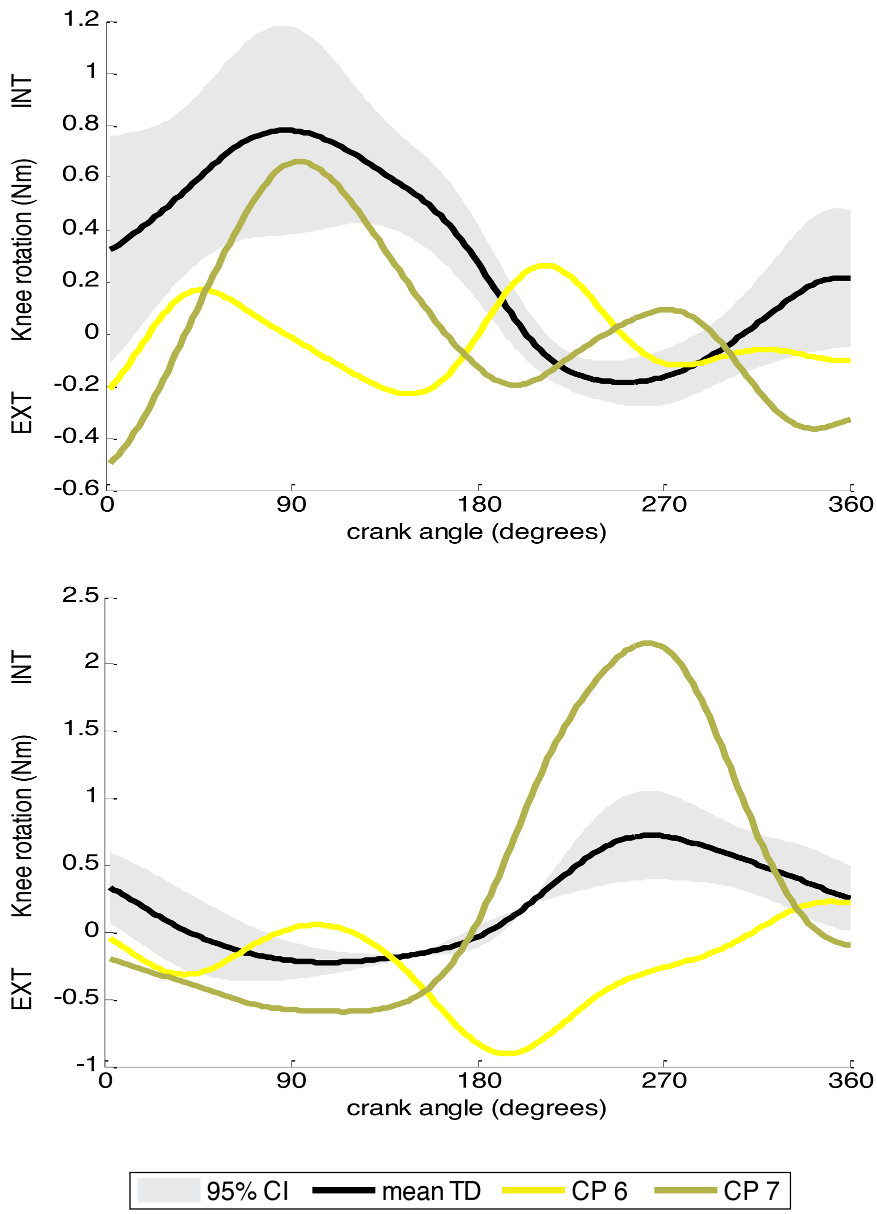


Figure A122: Knee rotation torques at 80 RPM produced by right (top) and left (bottom) leg.

ANKLE FLEXION TORQUES

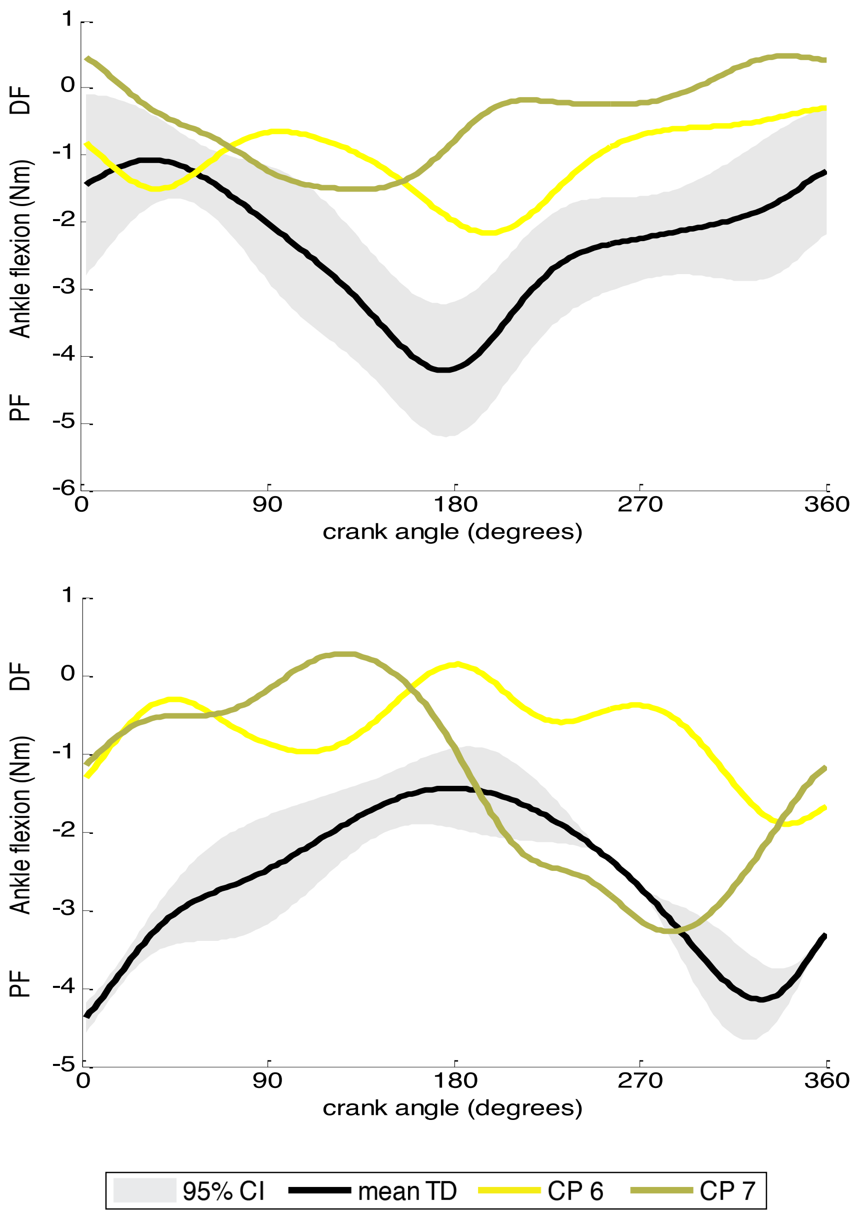


Figure A123: Ankle flexion torques at 80 RPM produced by right (top) and left (bottom) leg.

SUBTALAR ROTATION TORQUES

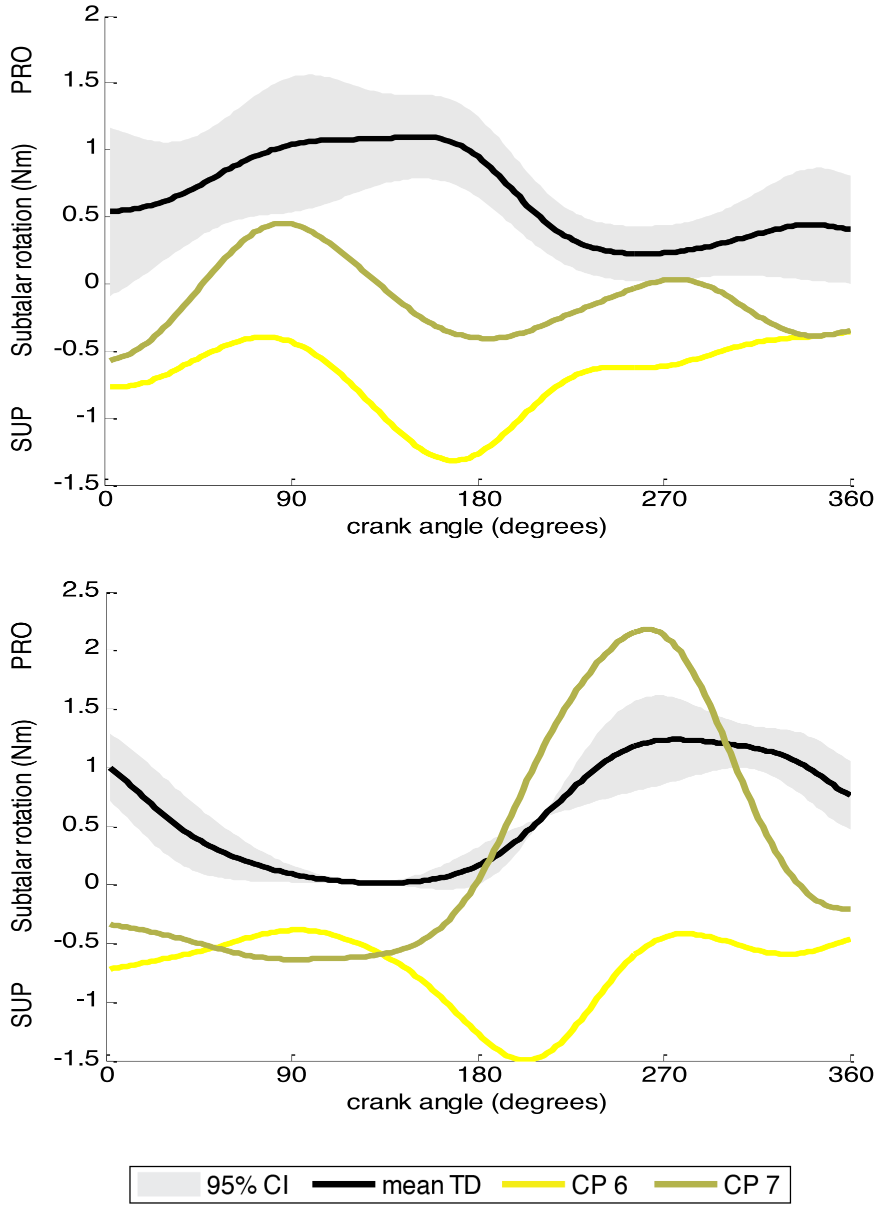
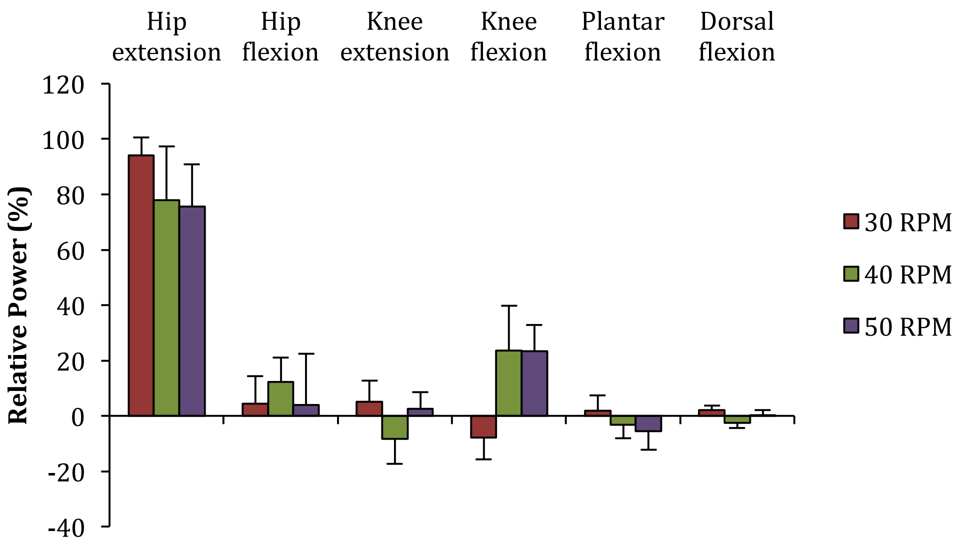


Figure A124: Subtalar rotation torques at 80 RPM produced by right (top) and left (bottom) leg.

APPENDIX AE: JOINT POWERS PER PARTICIPANT IN STUDY 2



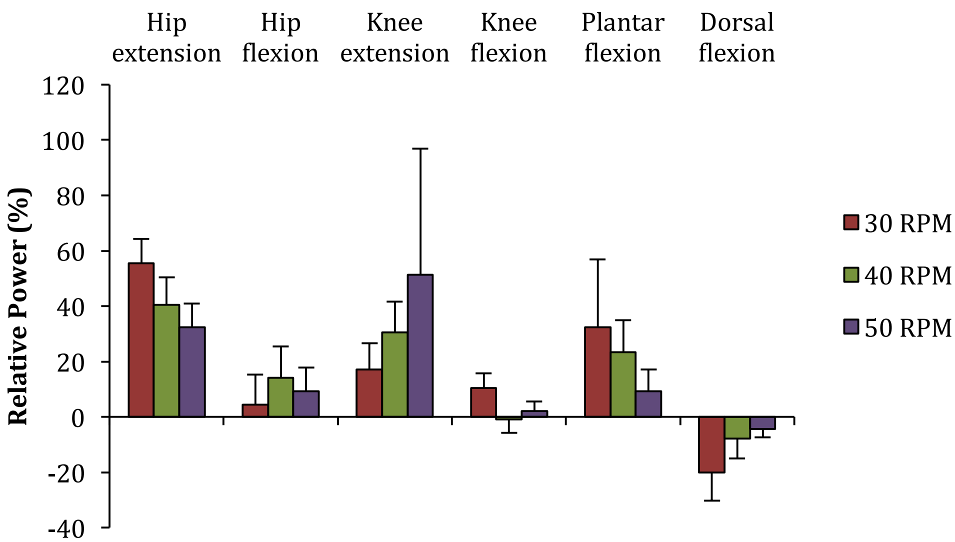


Figure A125: Joint powers produced by CP 1 in right (top) and left (bottom) leg.

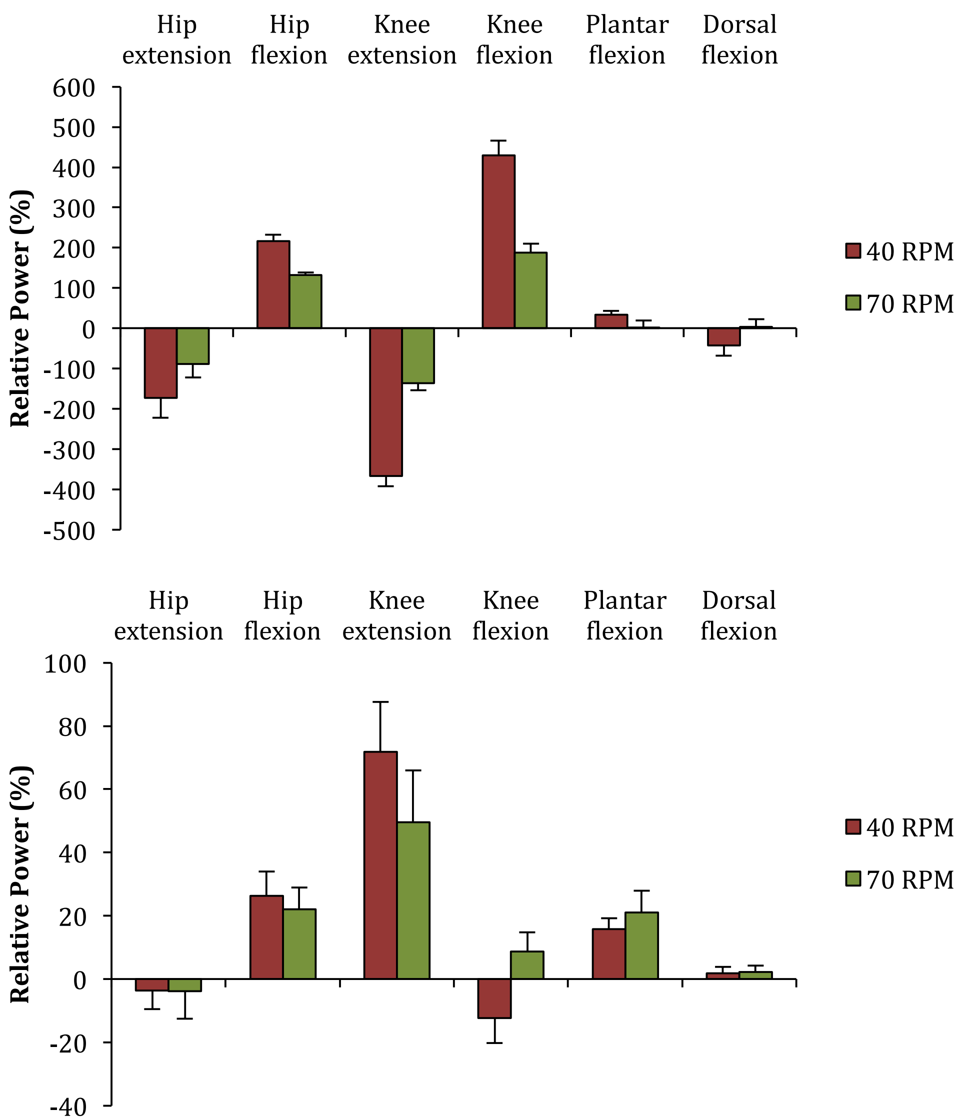


Figure A126: Joint powers produced by CP 2 in right (top) and left (bottom) leg.

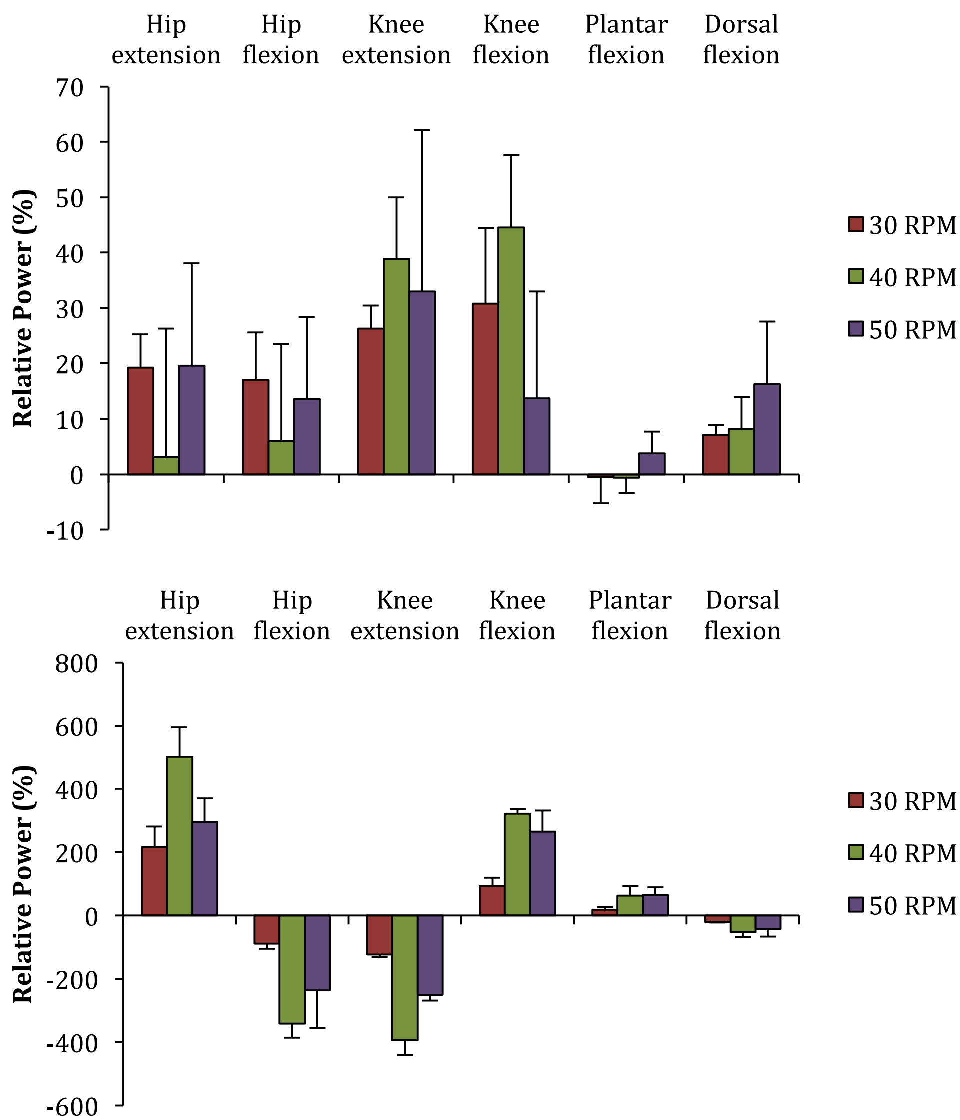


Figure A127: Joint powers produced by CP 3 in right (top) and left (bottom) leg.

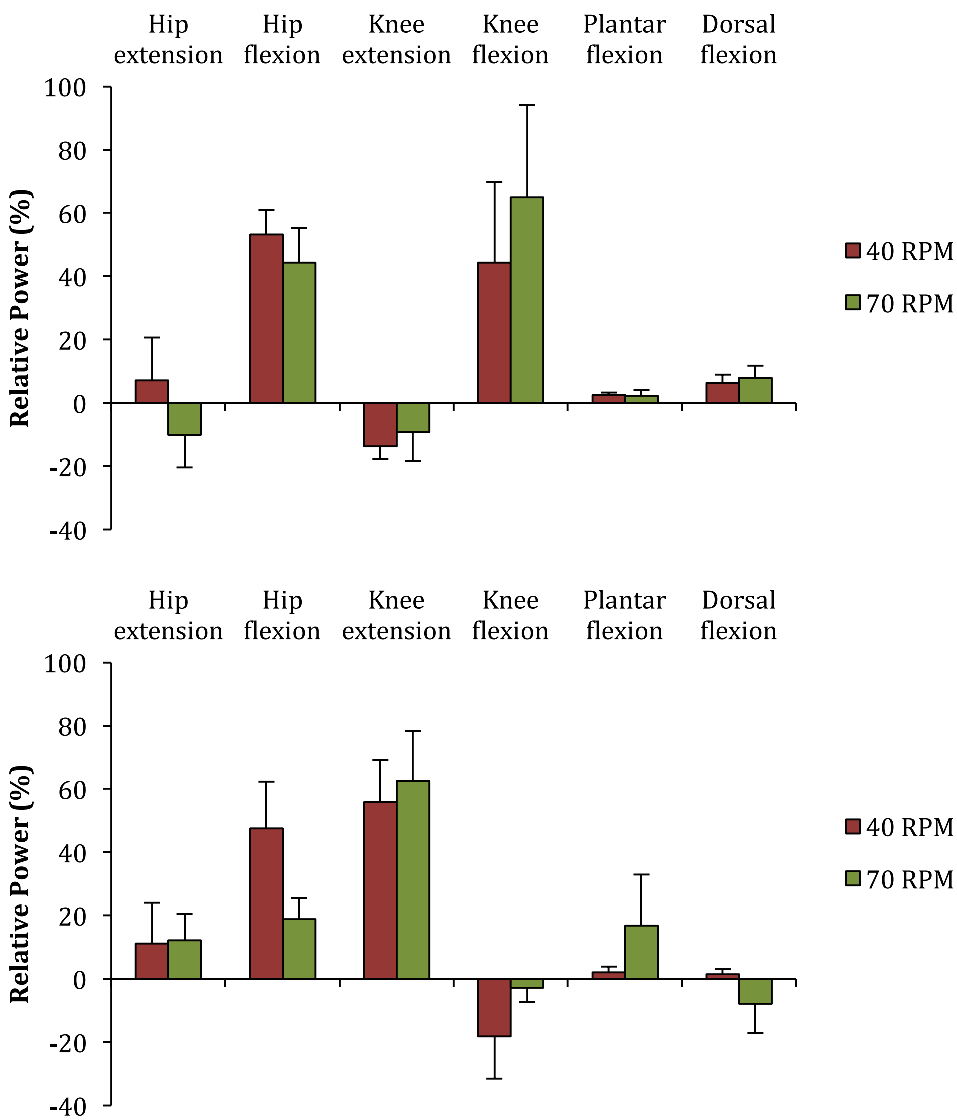


Figure A128: Joint powers produced by CP 5 in right (top) and left (bottom) leg.

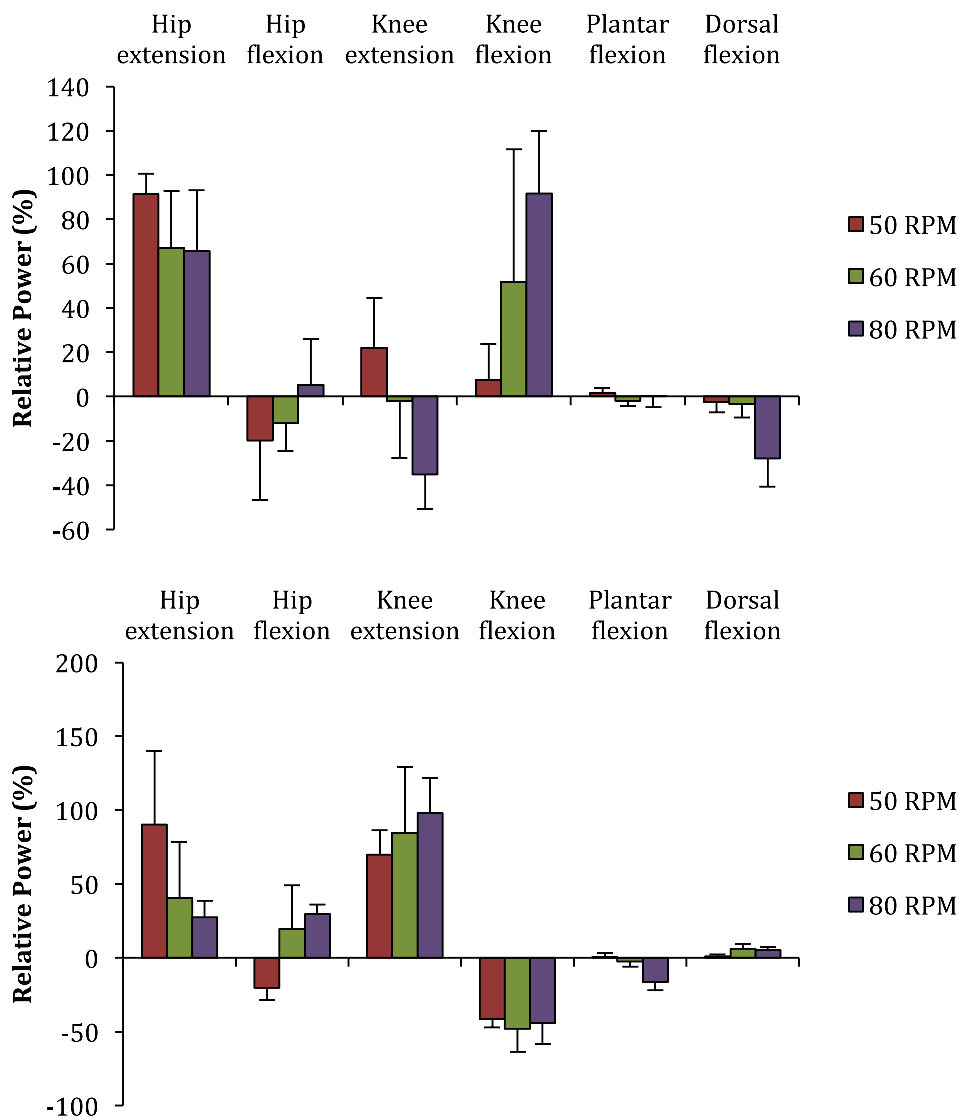


Figure A129: Joint powers produced by CP 6 in right (top) and left (bottom) leg.

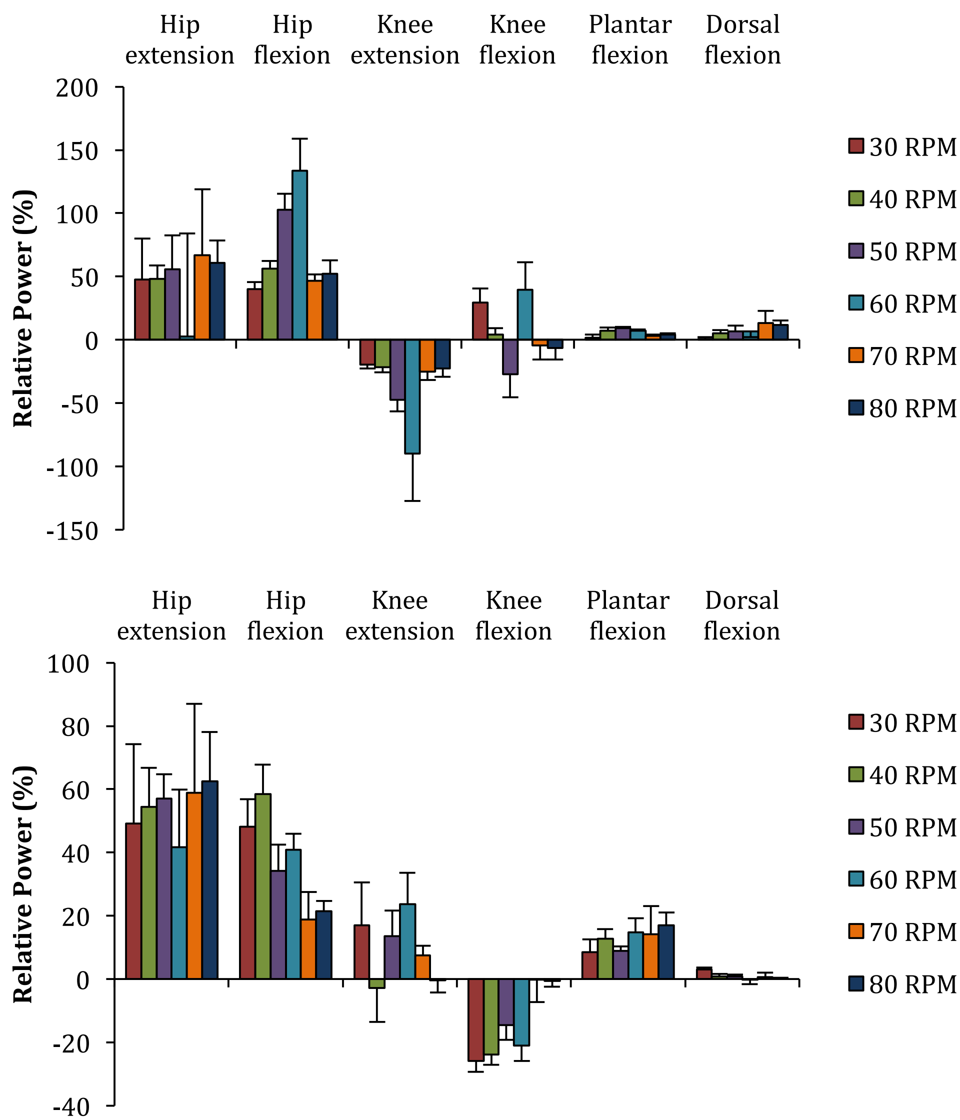


Figure A130: Joint powers produced by CP 7 in right (top) and left (bottom) leg.

APPENDIX AF: PHASE-PLANE DYNAMICS STUDY 3

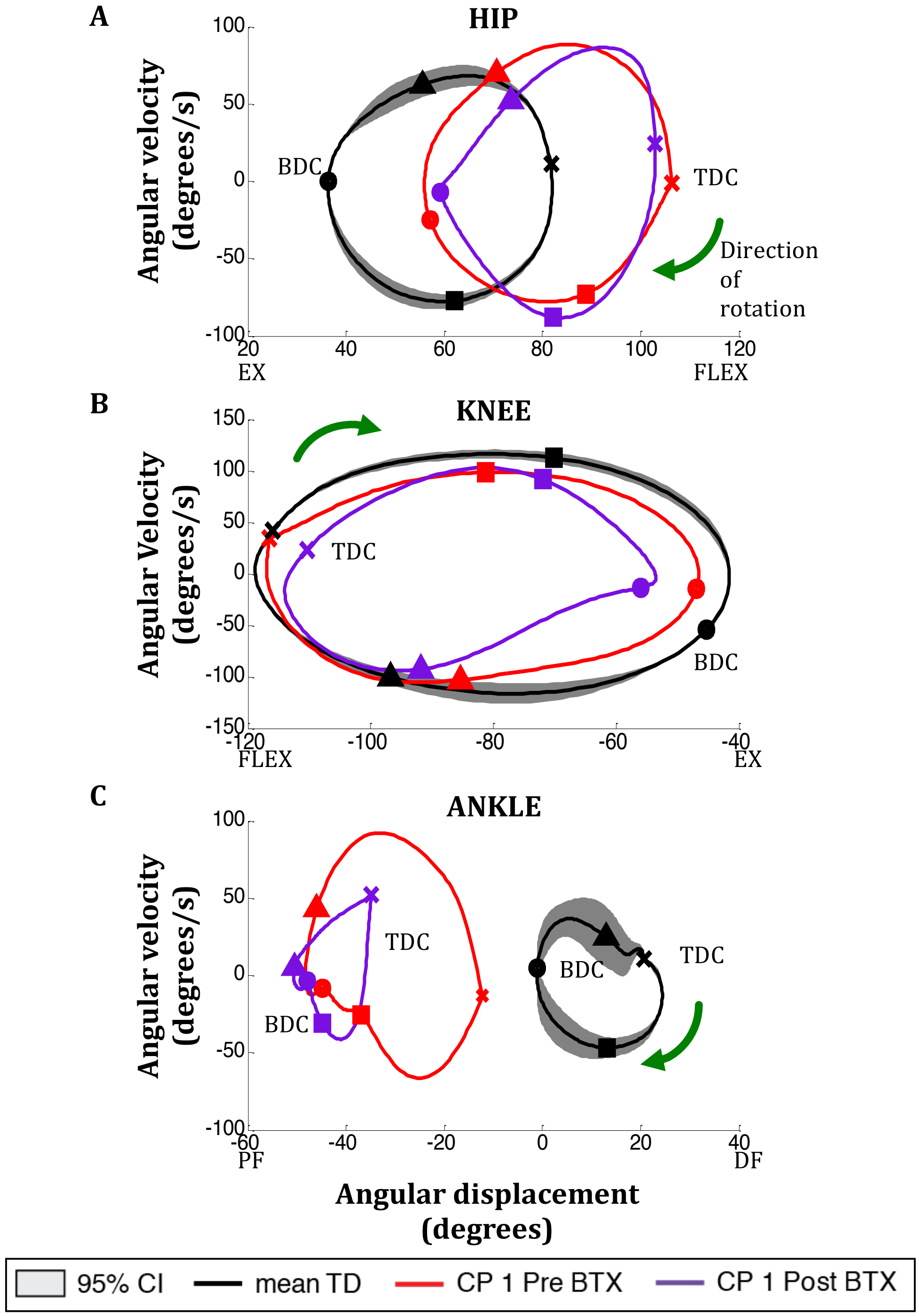


Figure A131: Phase-plane dynamics pre and post BTX at 30 RPM in treated leg.

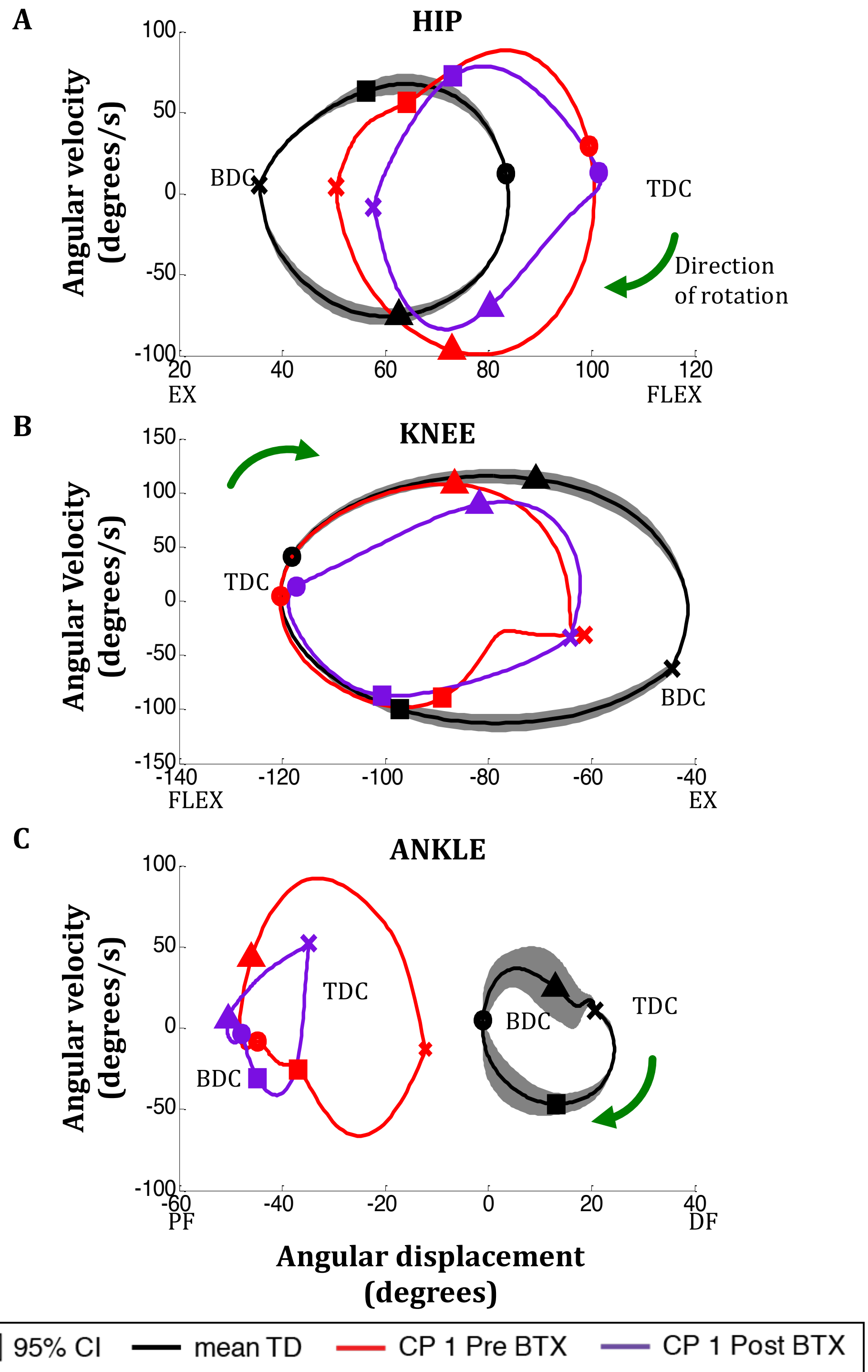


Figure A132: Phase-plane dynamics pre and post BTX at 30 RPM in non-treated leg.

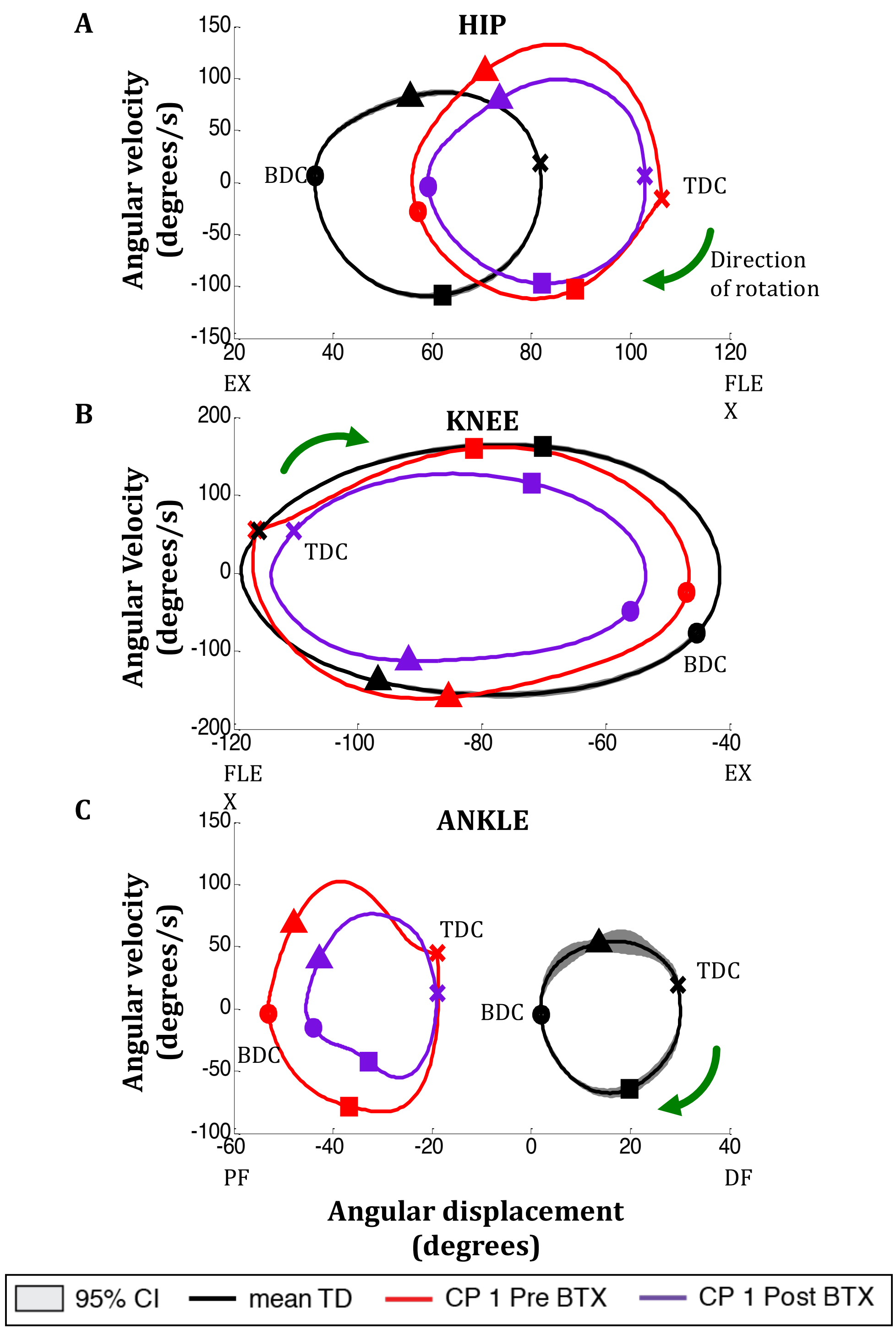


Figure A133: Phase-plane dynamics pre and post BTX at 40 RPM in treated leg.

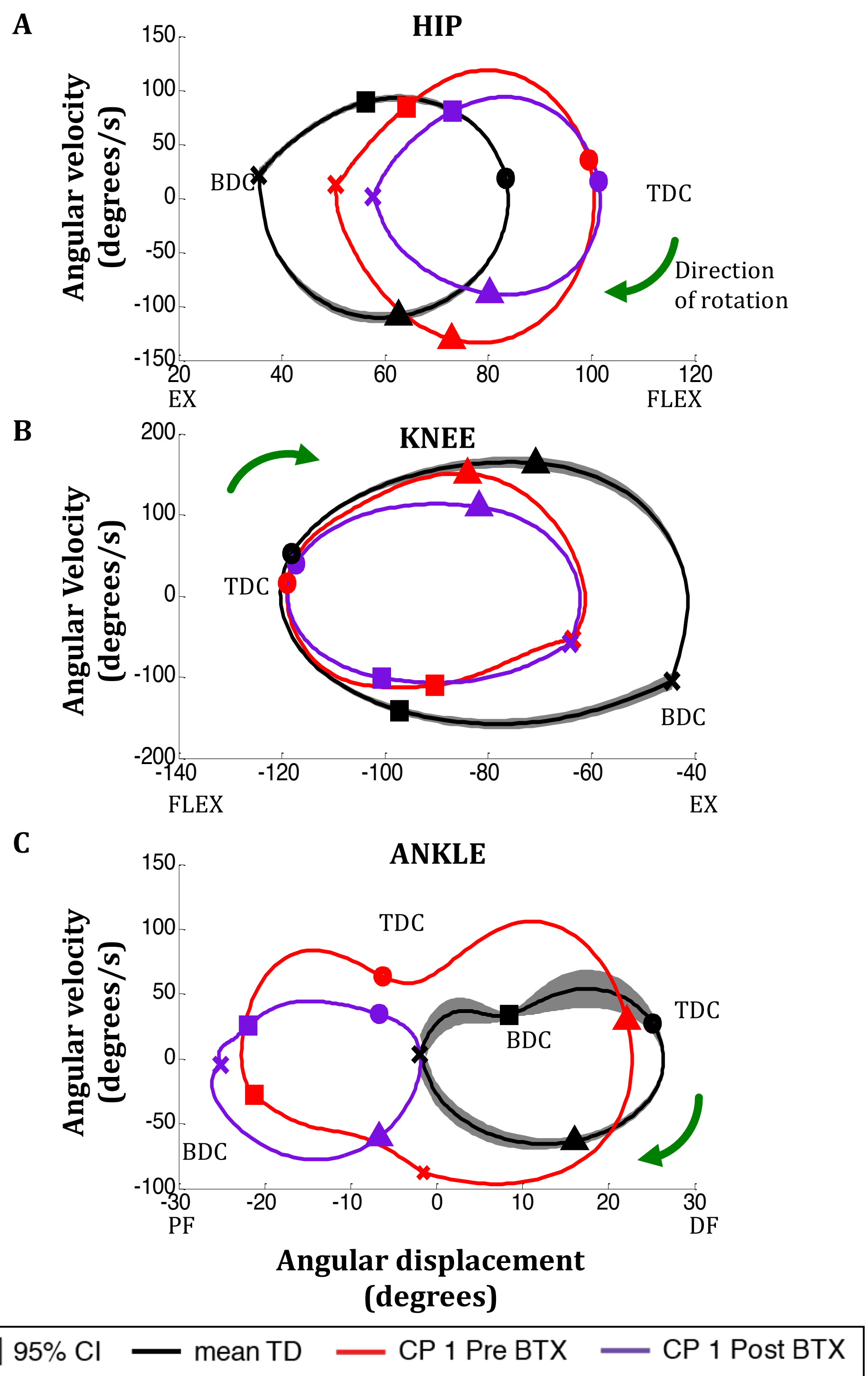


Figure A134: Phase-plane dynamics pre and post BTX at 40 RPM in non-treated leg.

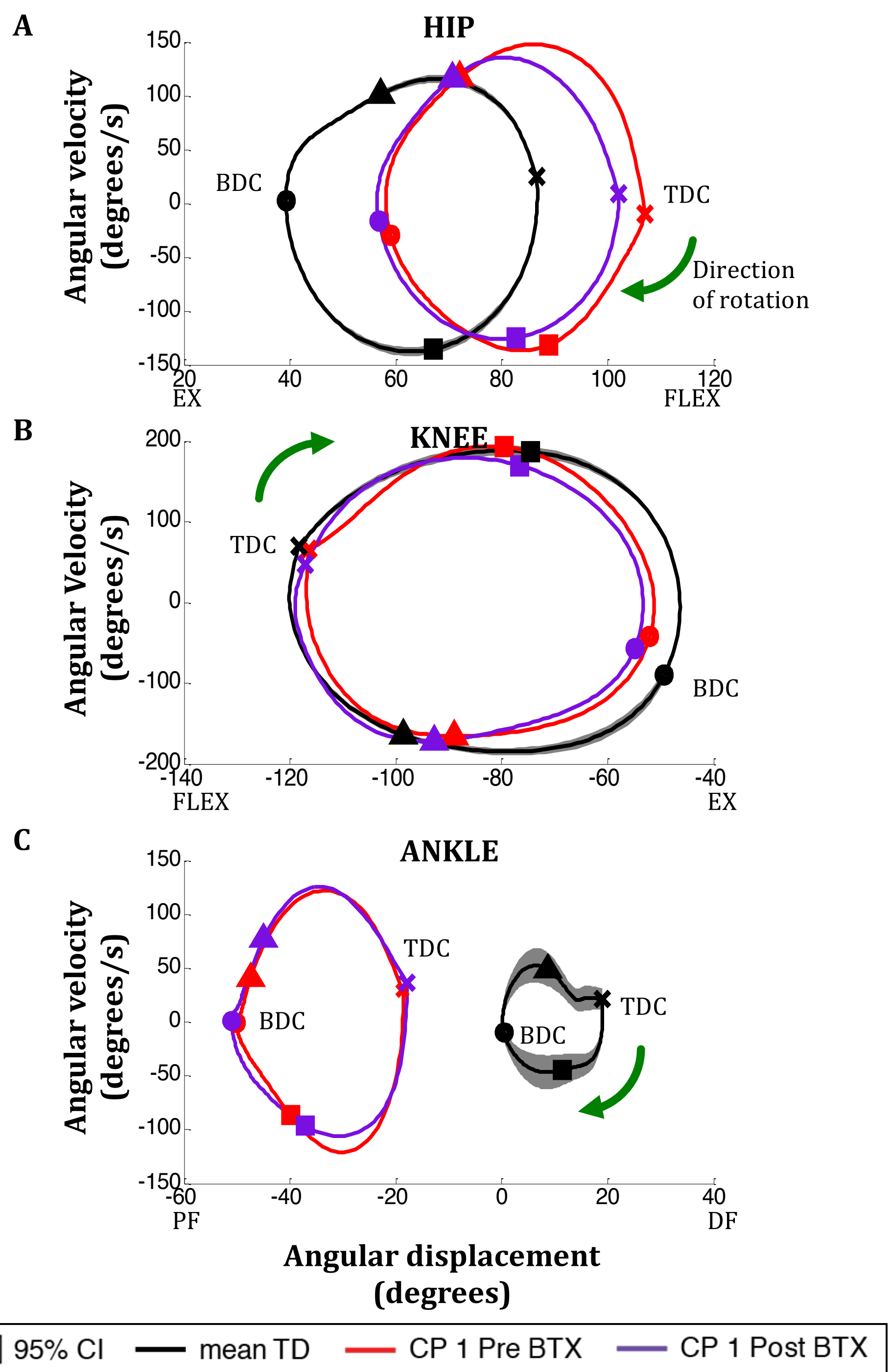


Figure A135: Phase-plane dynamics pre and post BTX at 50 RPM in treated leg.

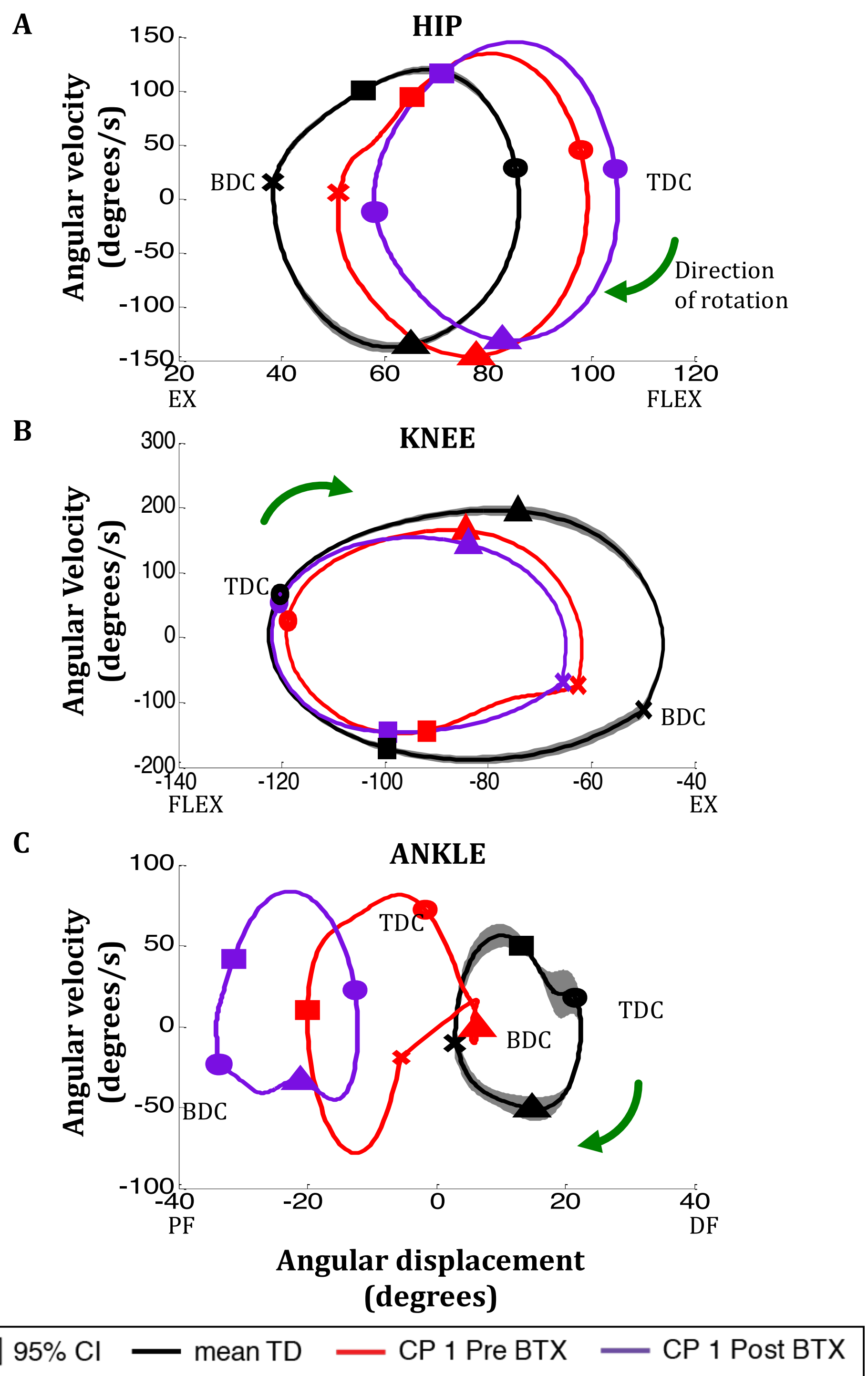


Figure A136: Phase-plane dynamics pre and post BTX at 50 RPM in non-treated leg.

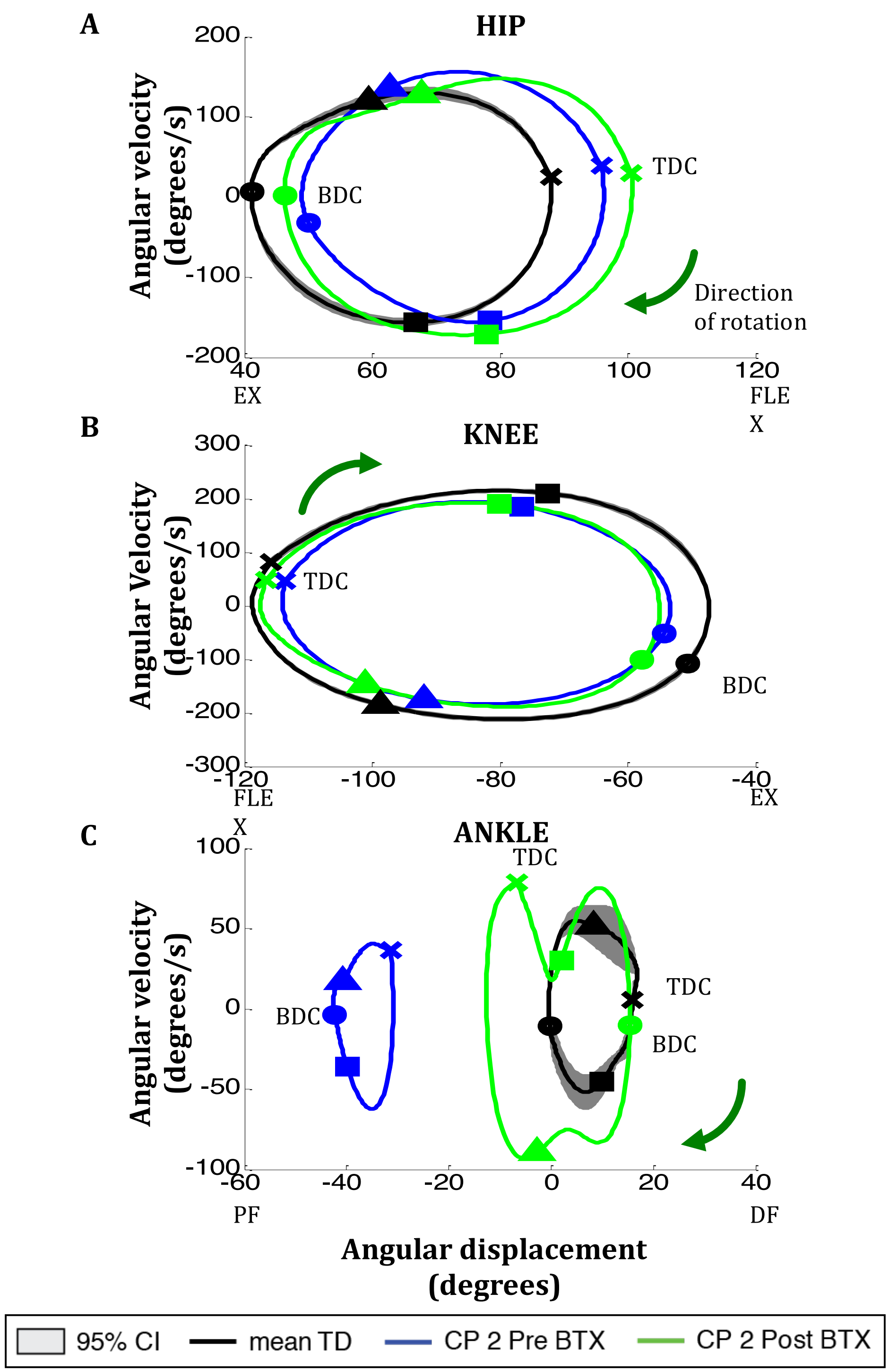


Figure A137: Phase-plane dynamics pre and post BTX at 60 RPM in treated leg.

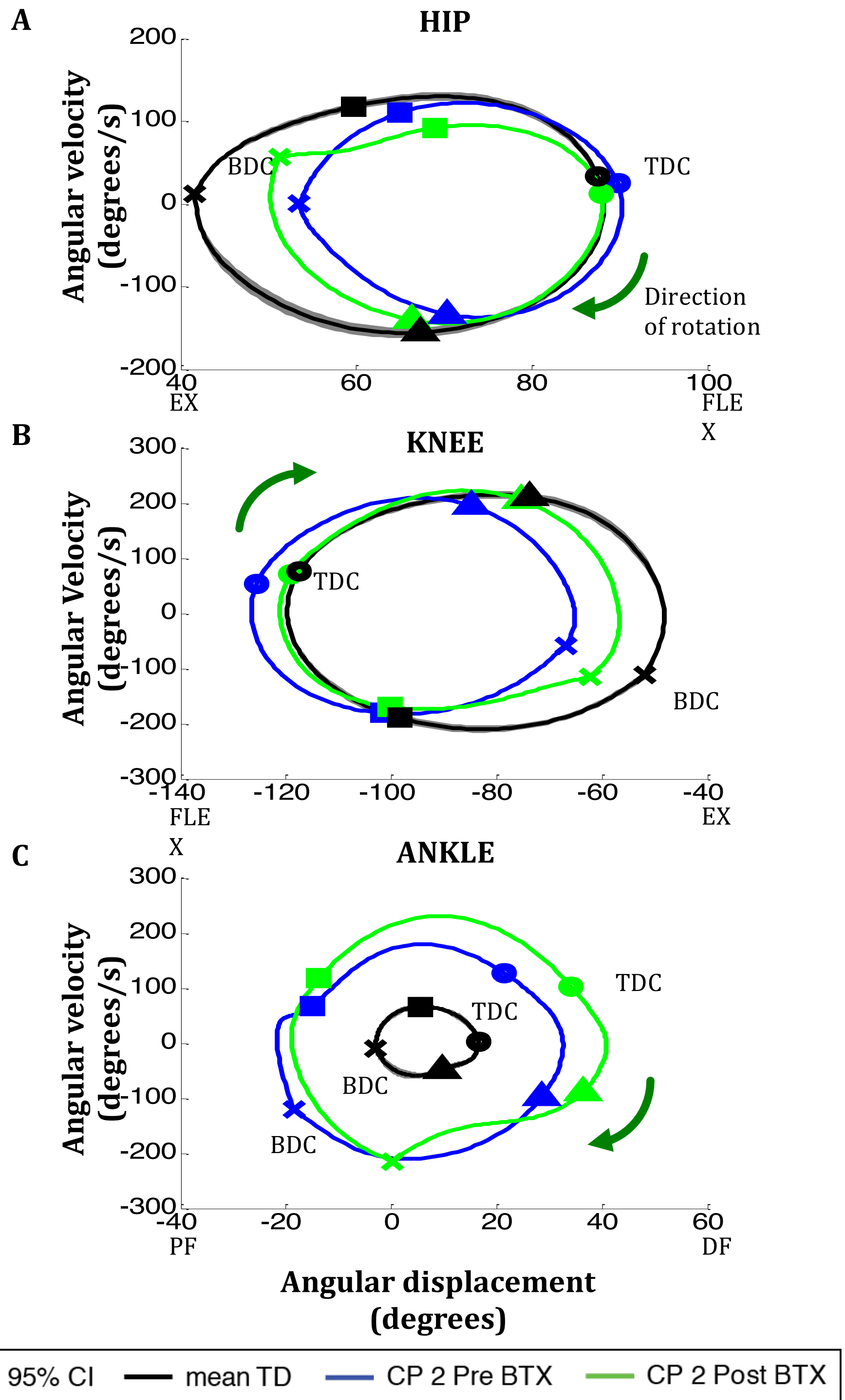


Figure A138: Phase-plane dynamics pre and post BTX at 60 RPM in non-treated leg.

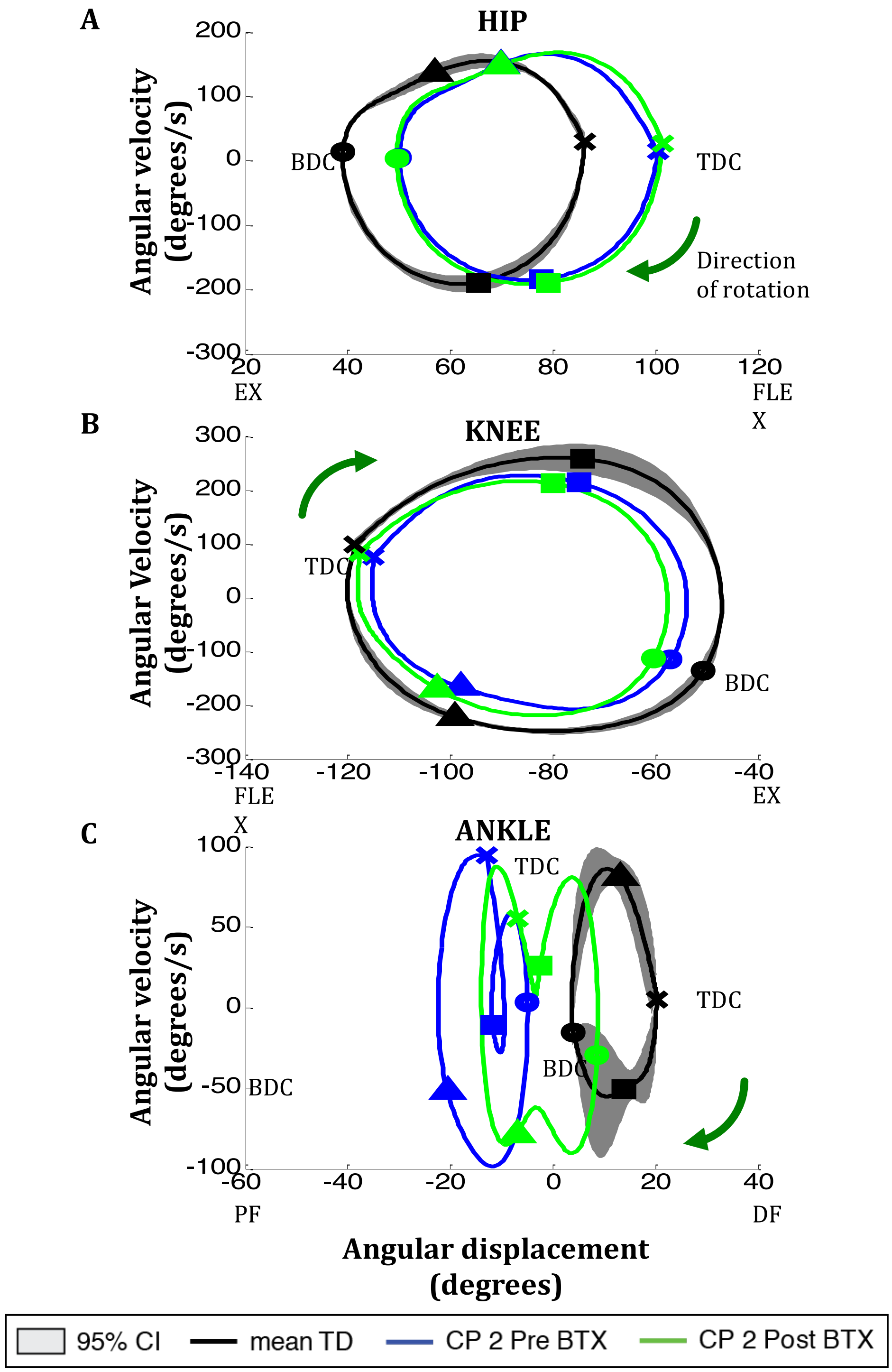


Figure A139: Phase-plane dynamics pre and post BTX at 70 RPM in treated leg.

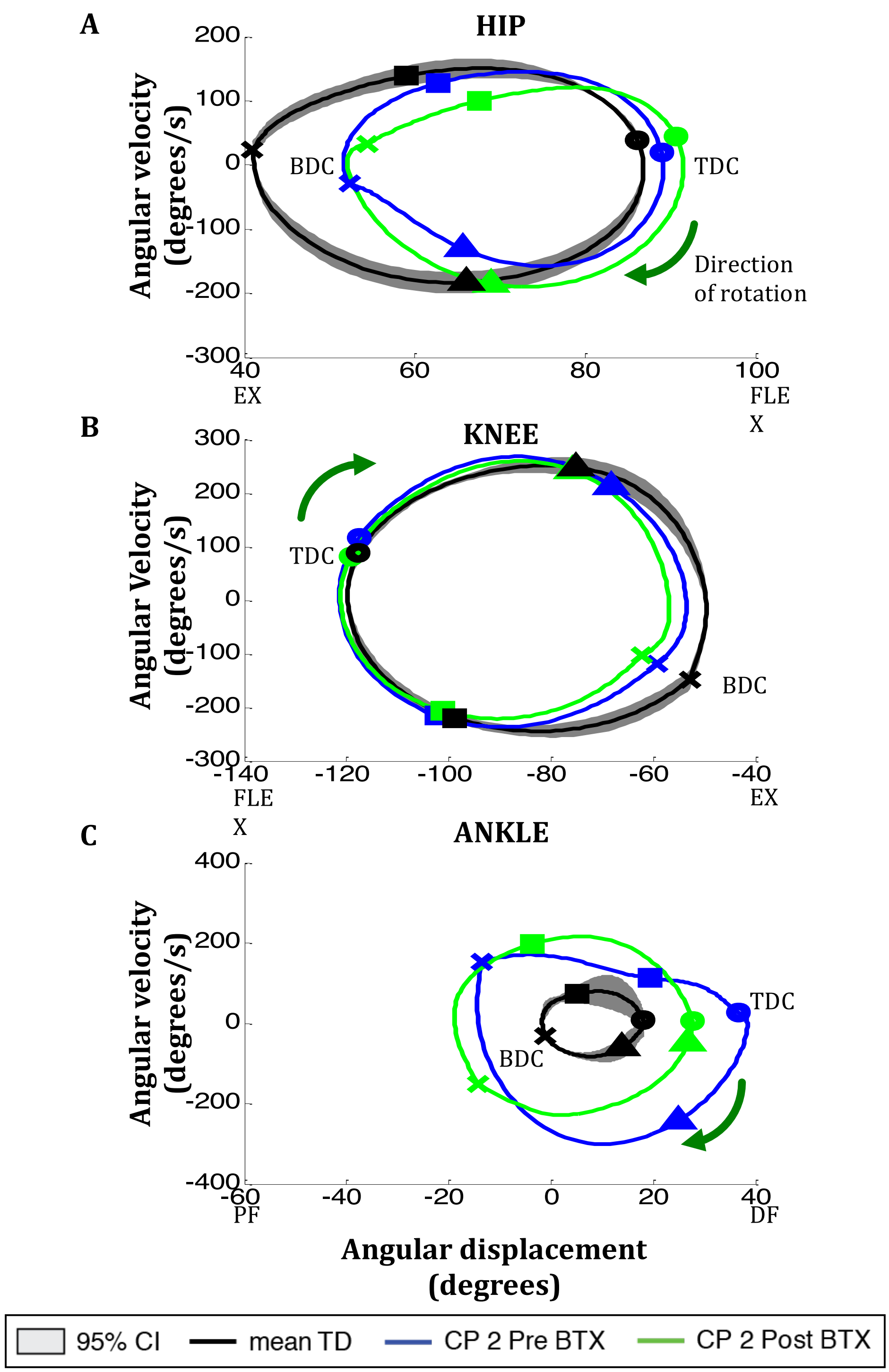


Figure A140: Phase-plane dynamics pre and post BTX at 70 RPM in non-treated leg.

APPENDIX AG: JOINT TORQUES PARTICIPANT 1 AT 30 RPM IN STUDY 3

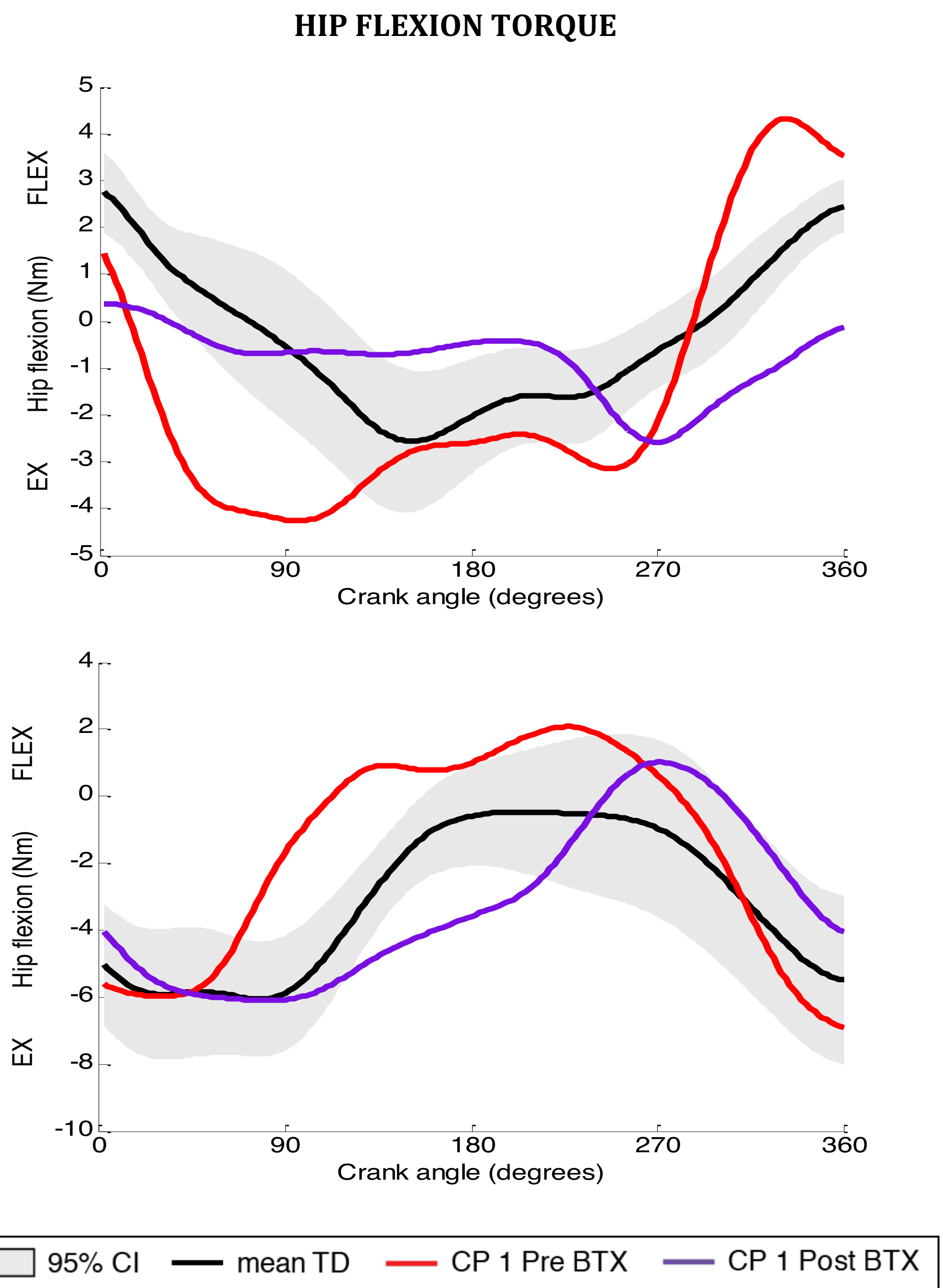


Figure A141: Hip flexion torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

HIP ADDUCTION TORQUE

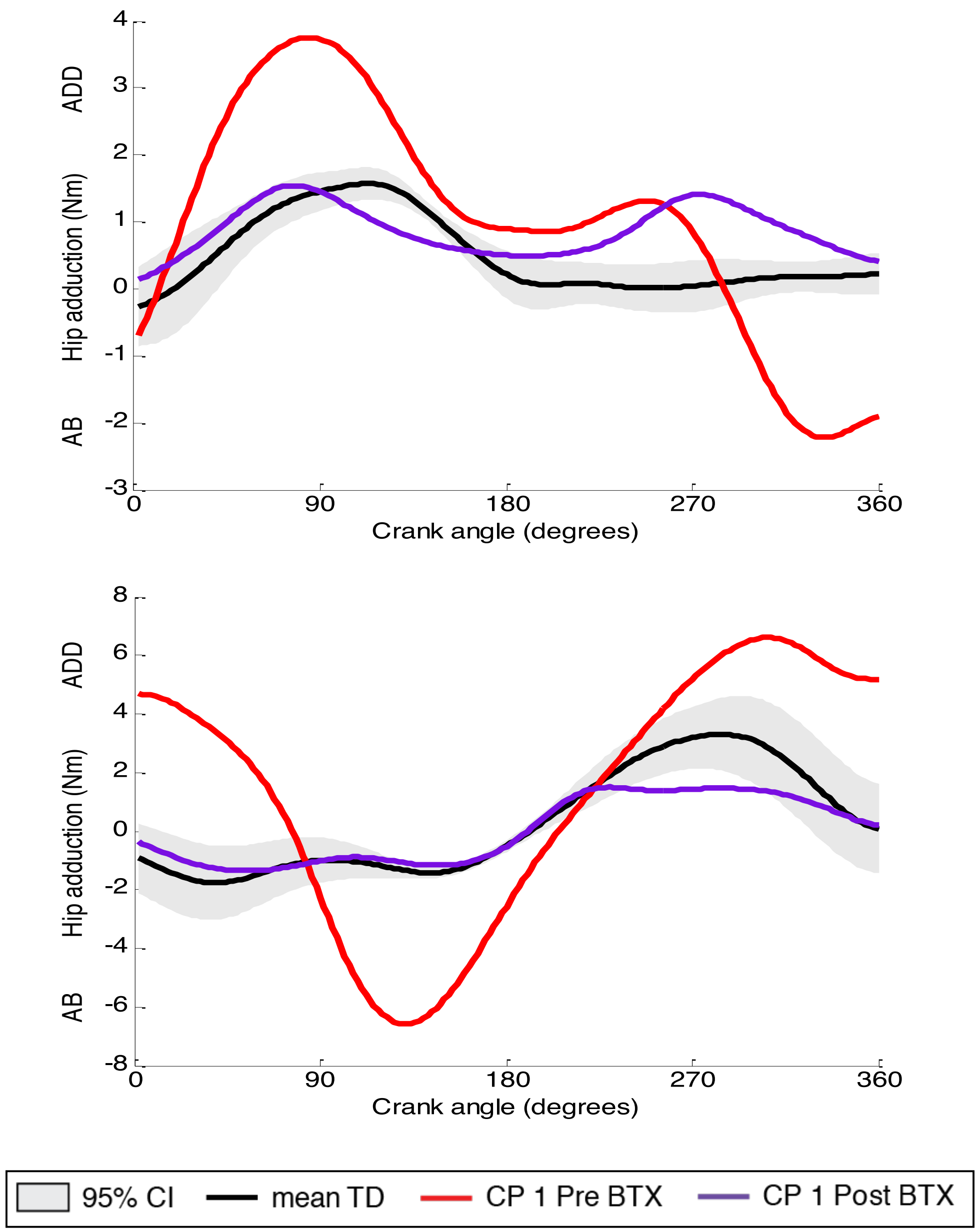


Figure A142: Hip adduction torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

HIP ROTATION TORQUE

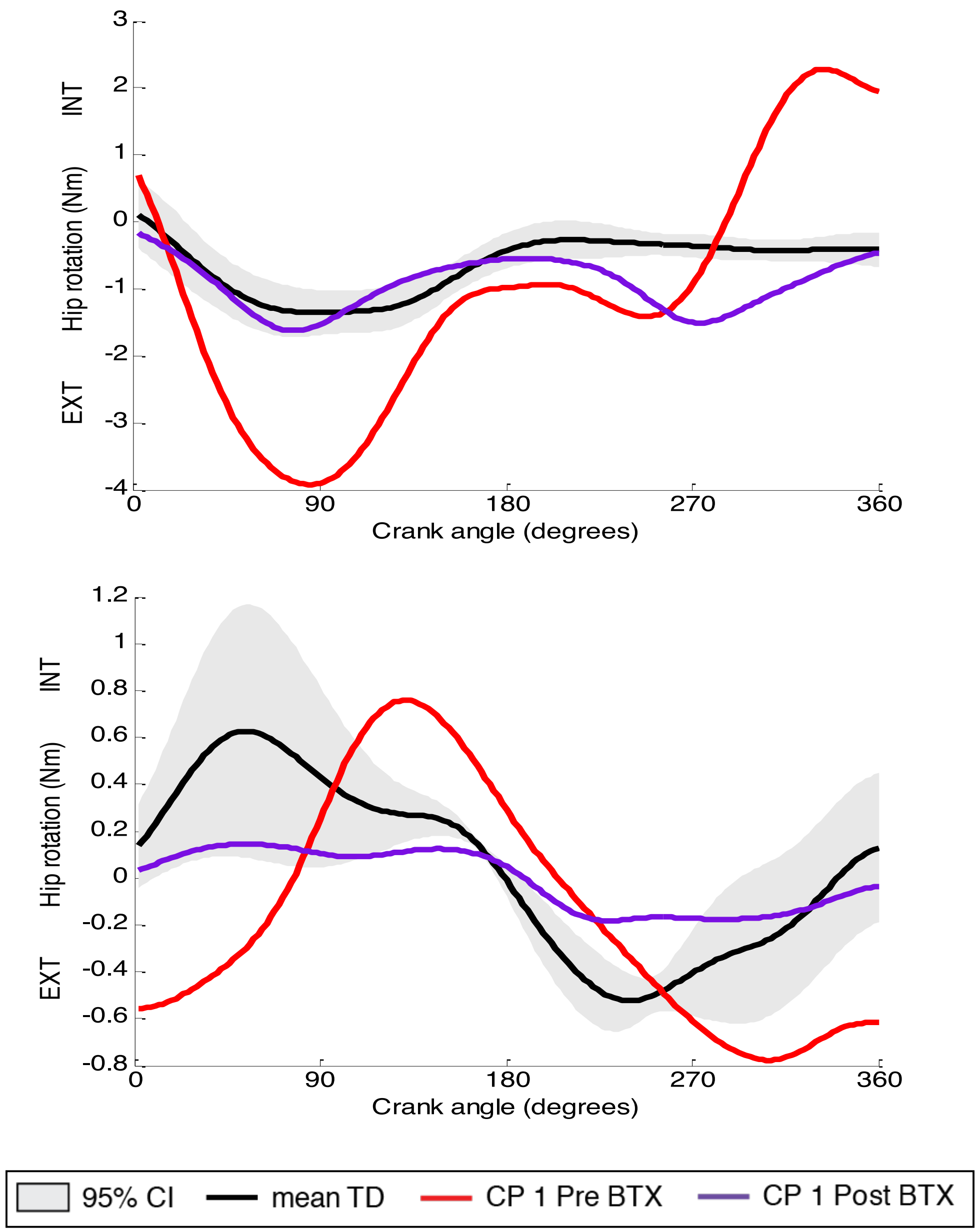


Figure A143: Hip rotation torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

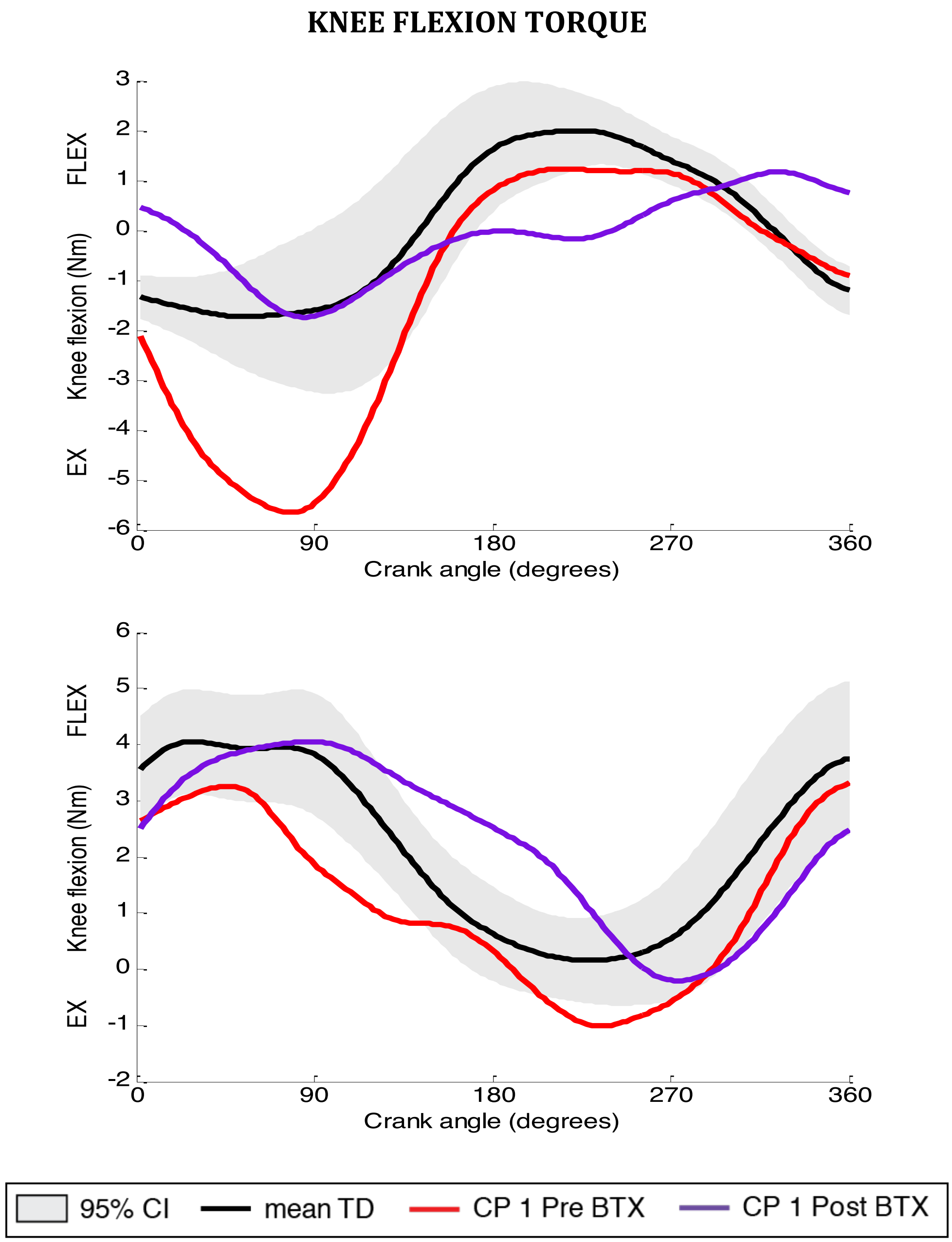


Figure A144: Knee flexion torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

KNEE ADDUCTION TORQUE

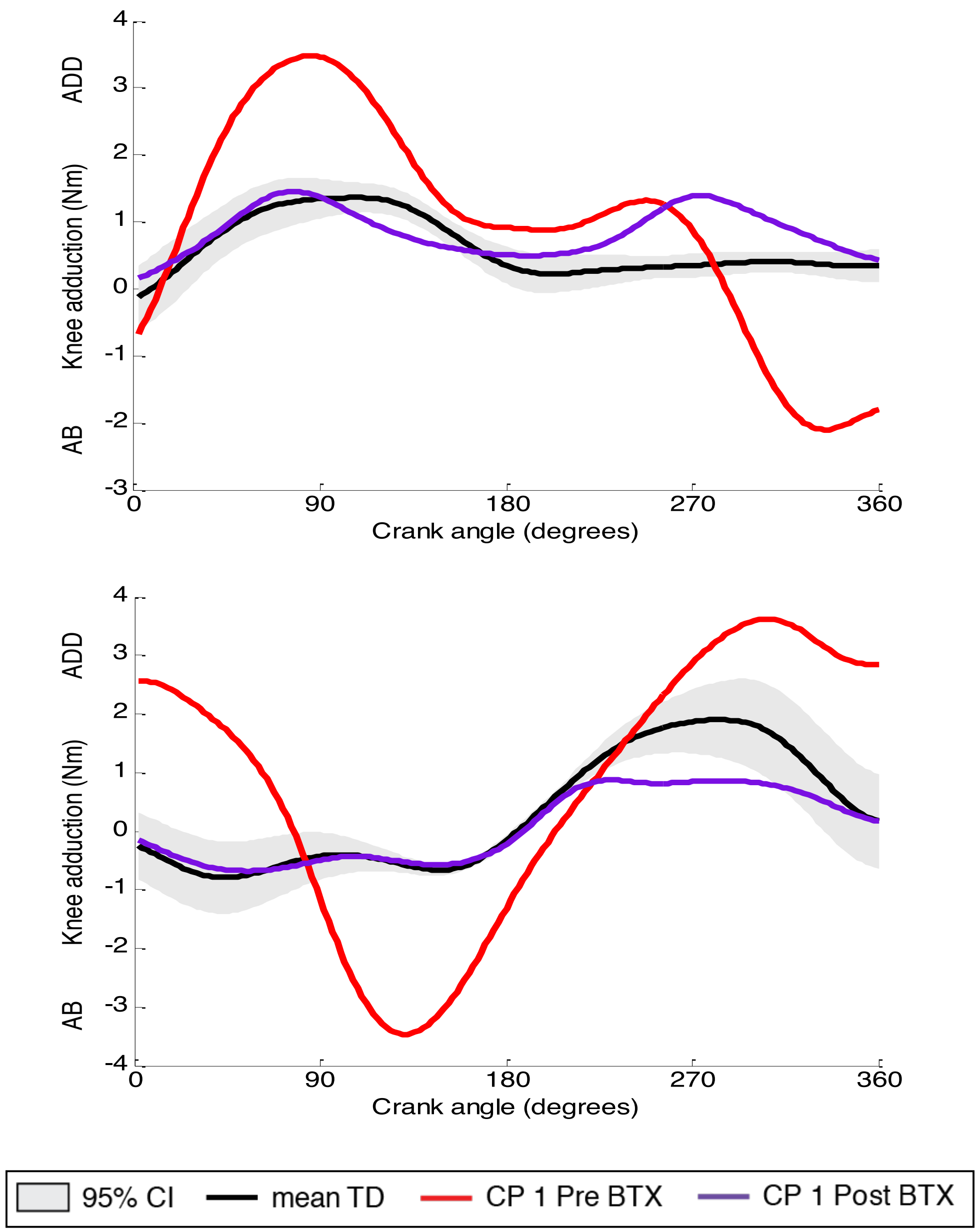


Figure A145: Knee adduction torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

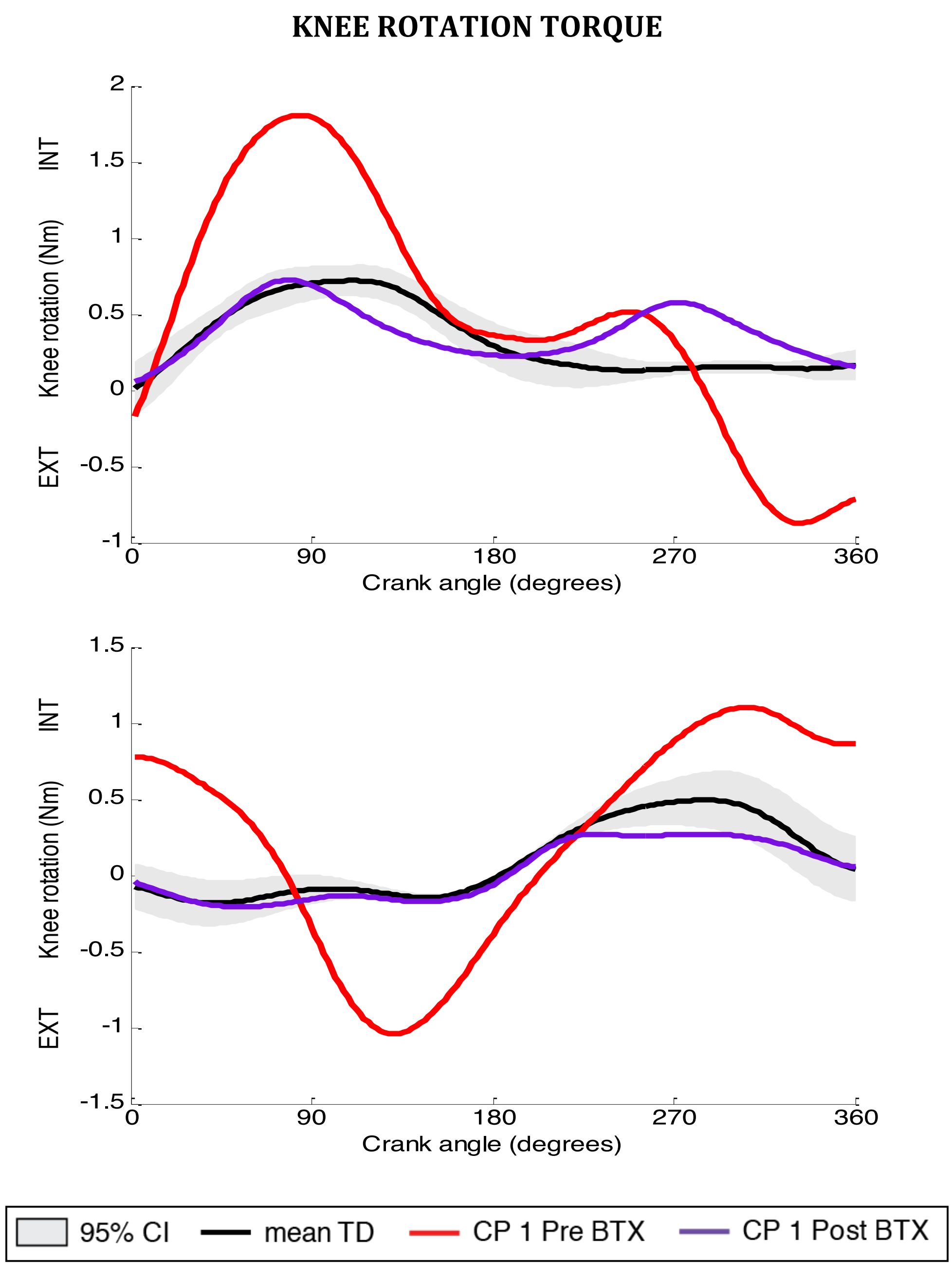


Figure A146: Knee rotation torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

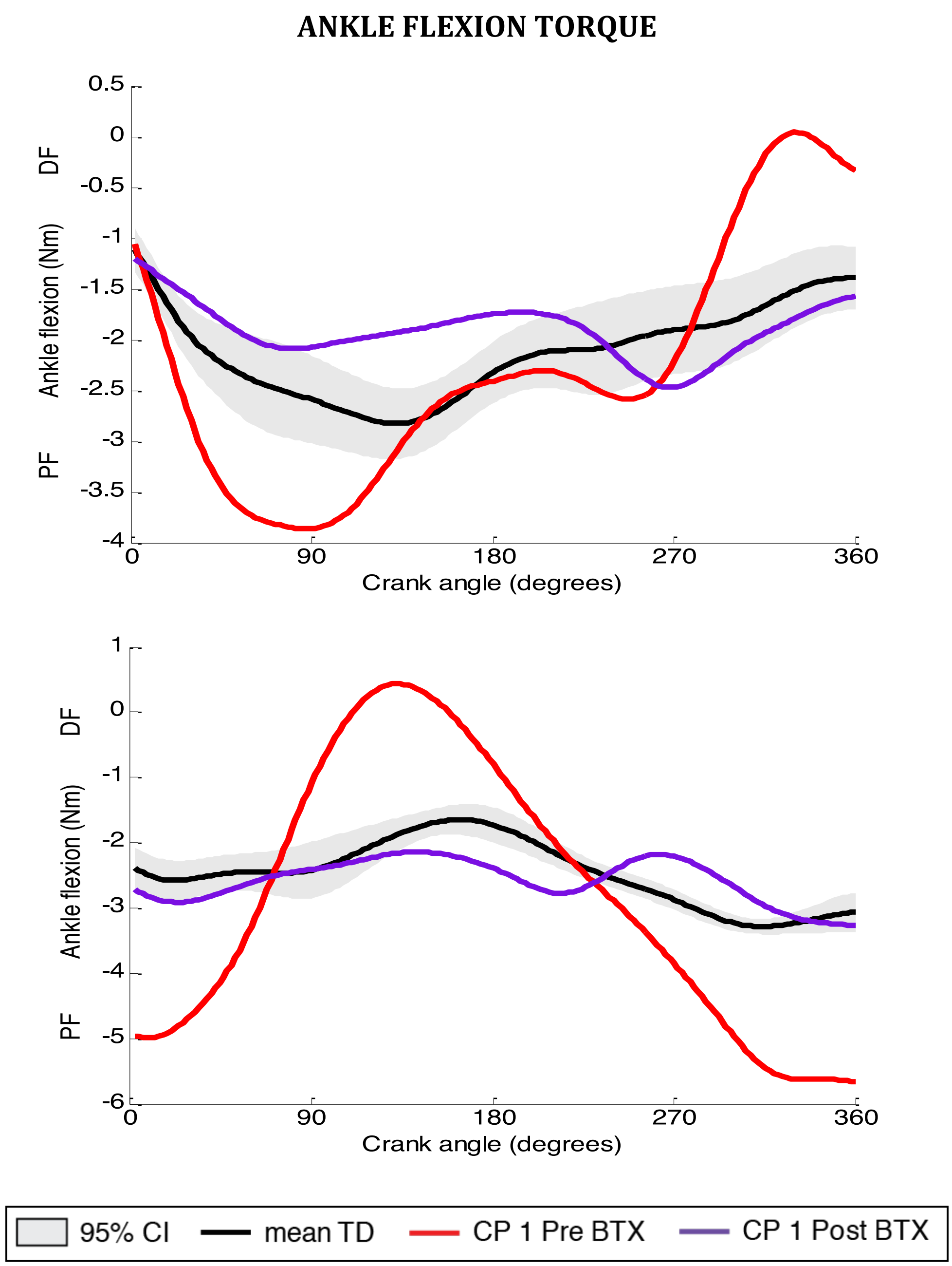


Figure A147: Ankle flexion torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

SUBTALAR ROTATION TORQUE

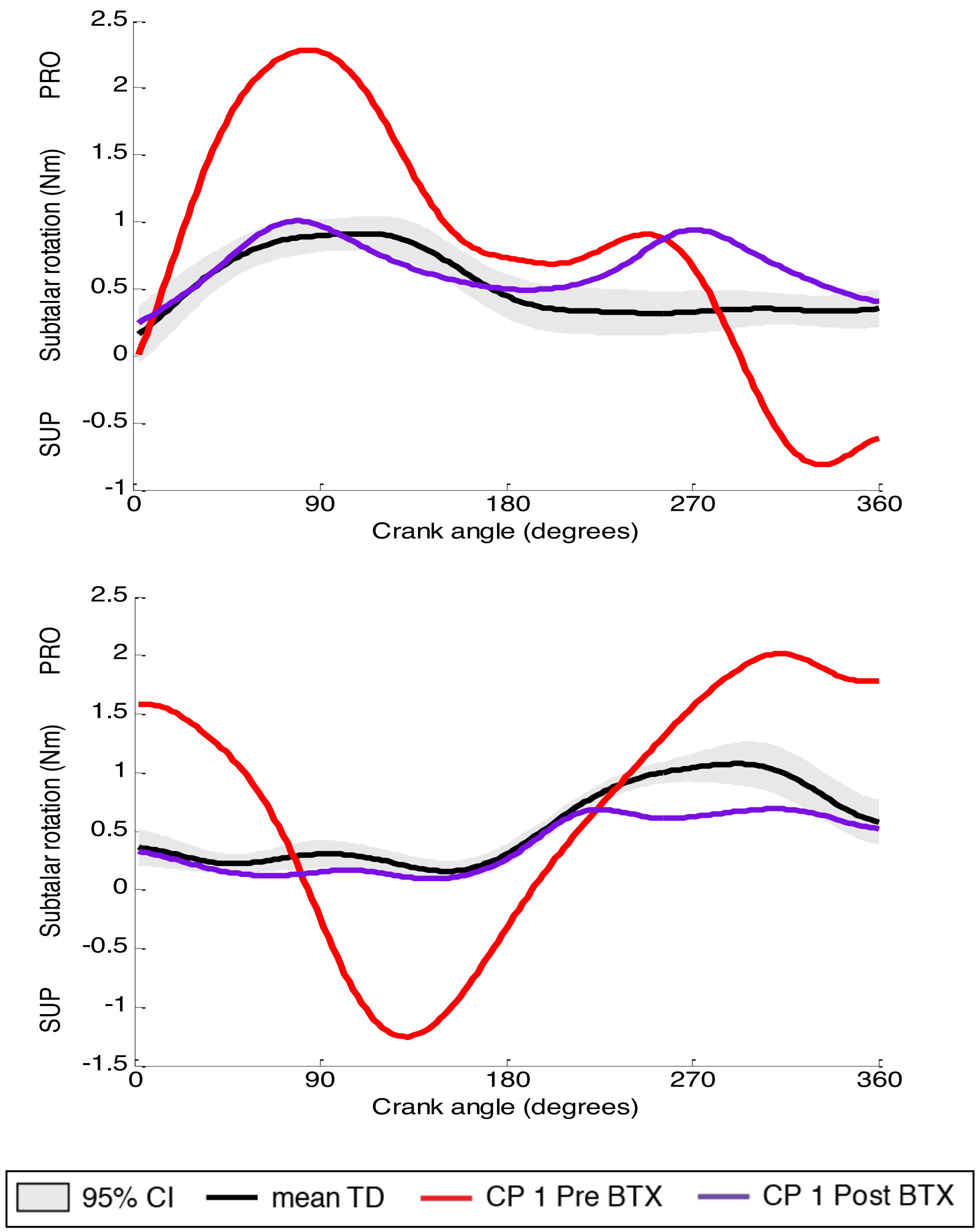


Figure A148: Subtalar rotation torques pre and post BTX at 30 RPM at treated (top) and non-treated (bottom) leg.

APPENDIX AH: JOINT TORQUES PARTICIPANT 1 AT 40 RPM IN STUDY 3

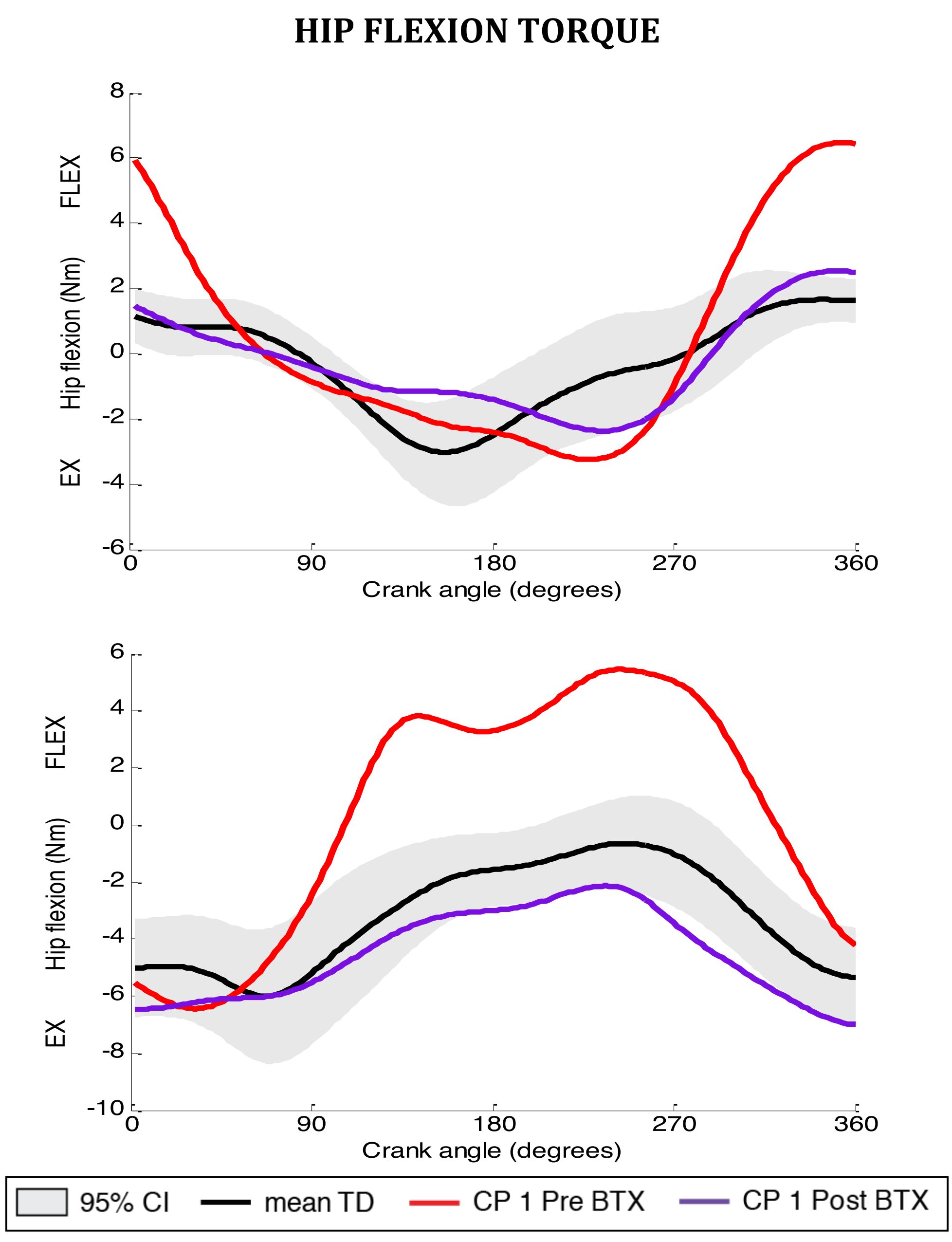


Figure A149: Hip flexion torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

HIP ADDUCTION TORQUE

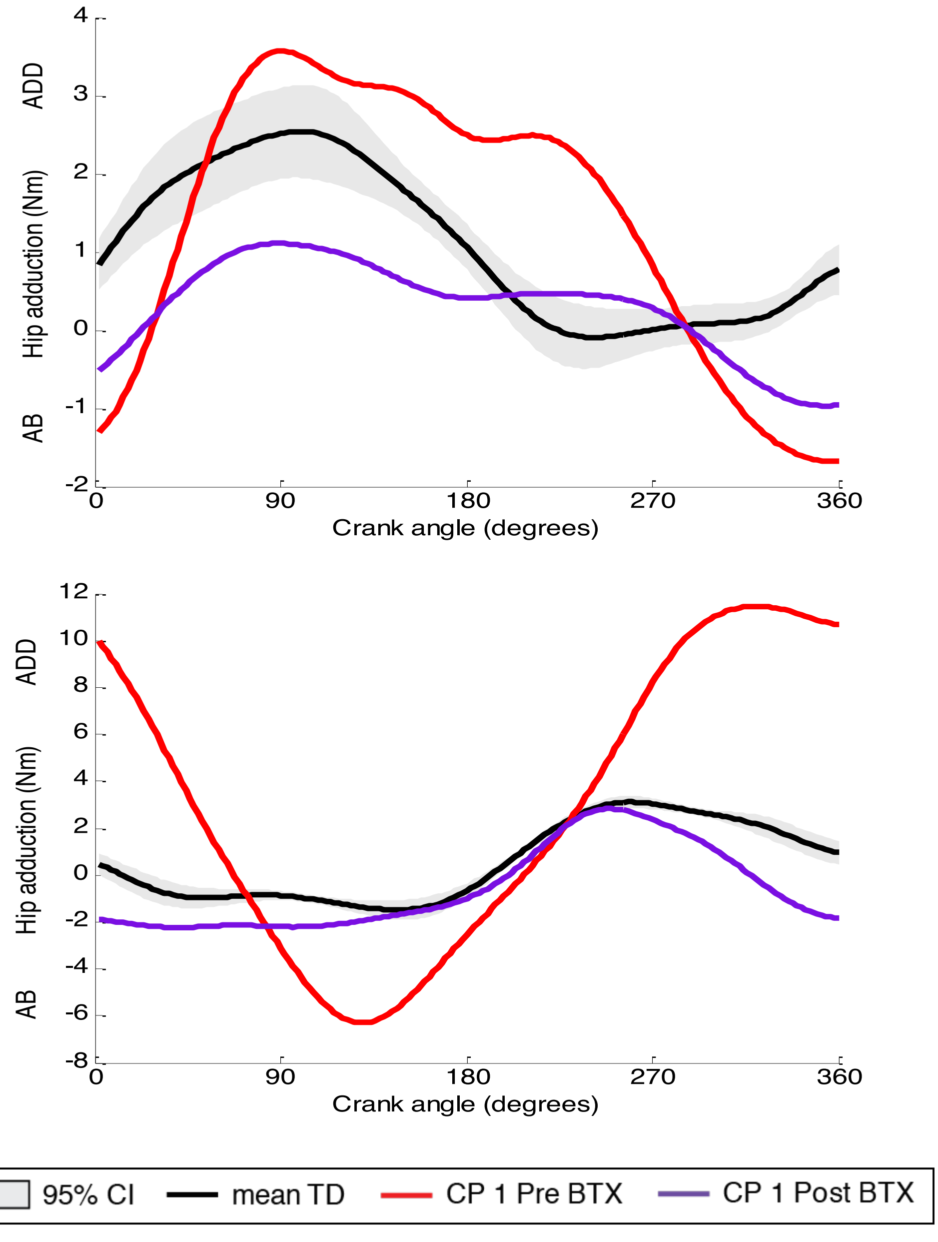


Figure A150: Hip adduction torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

HIP ROTATION TORQUE

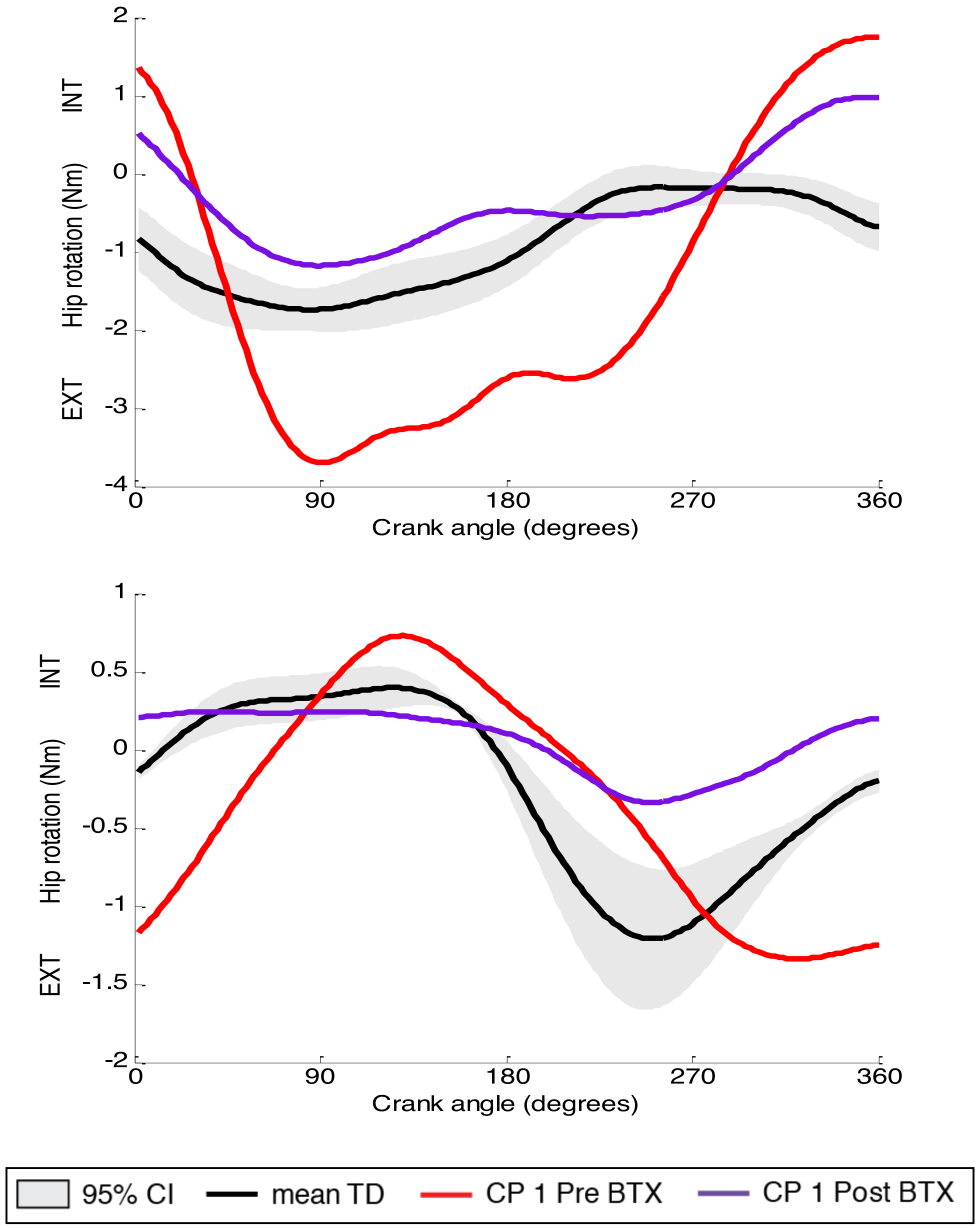


Figure A151: Hip rotation torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

KNEE FLEXION TORQUE

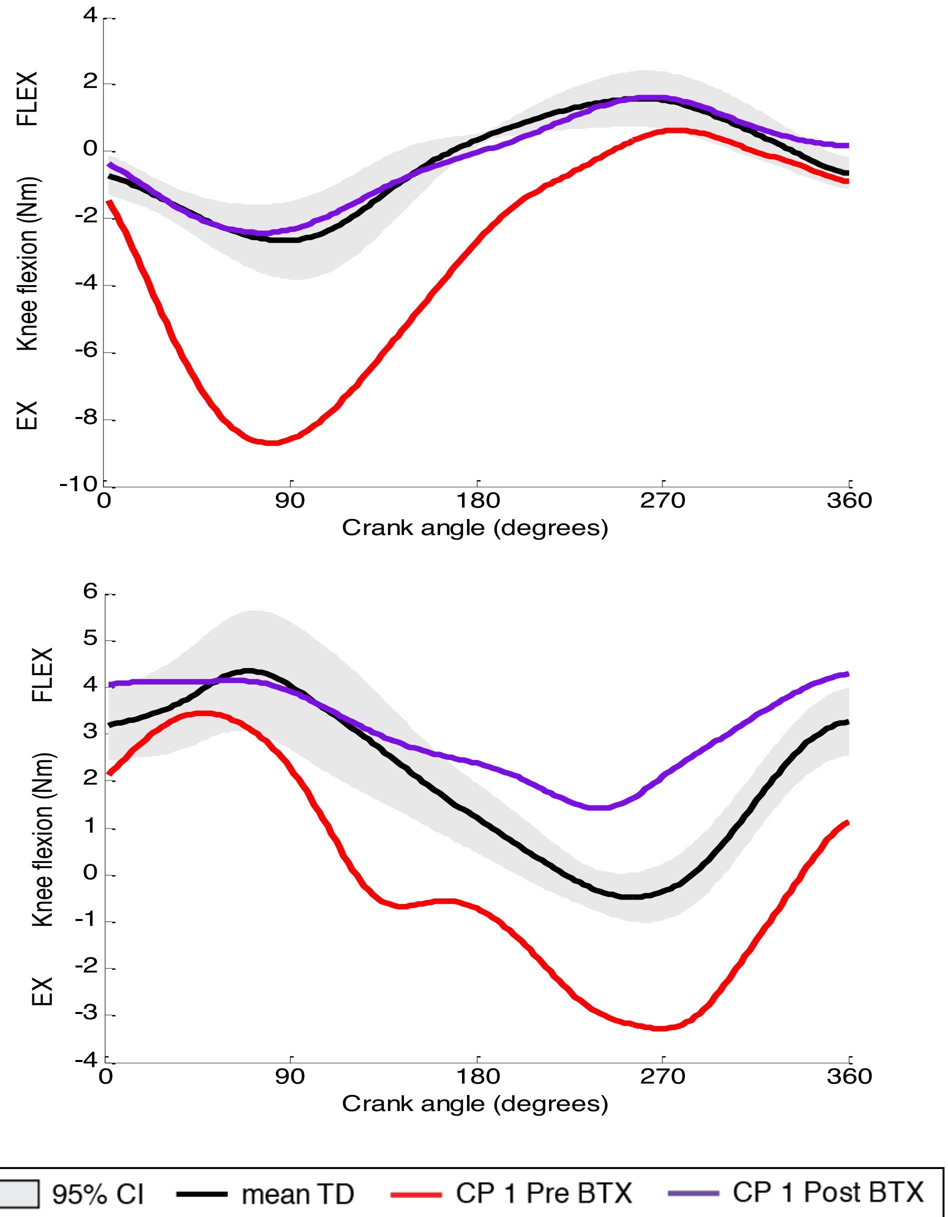


Figure A152: Knee flexion torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

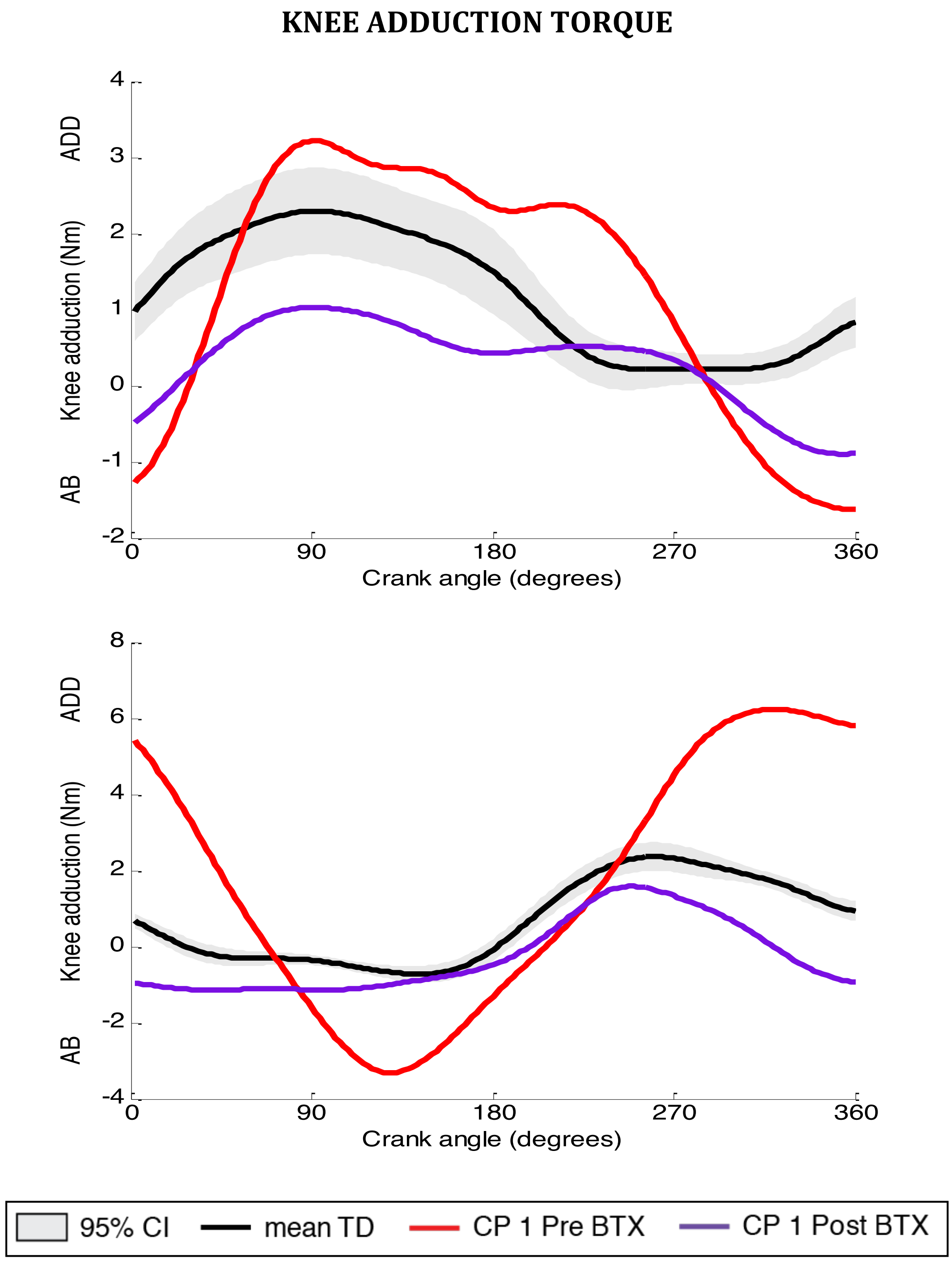


Figure A153: Knee adduction torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

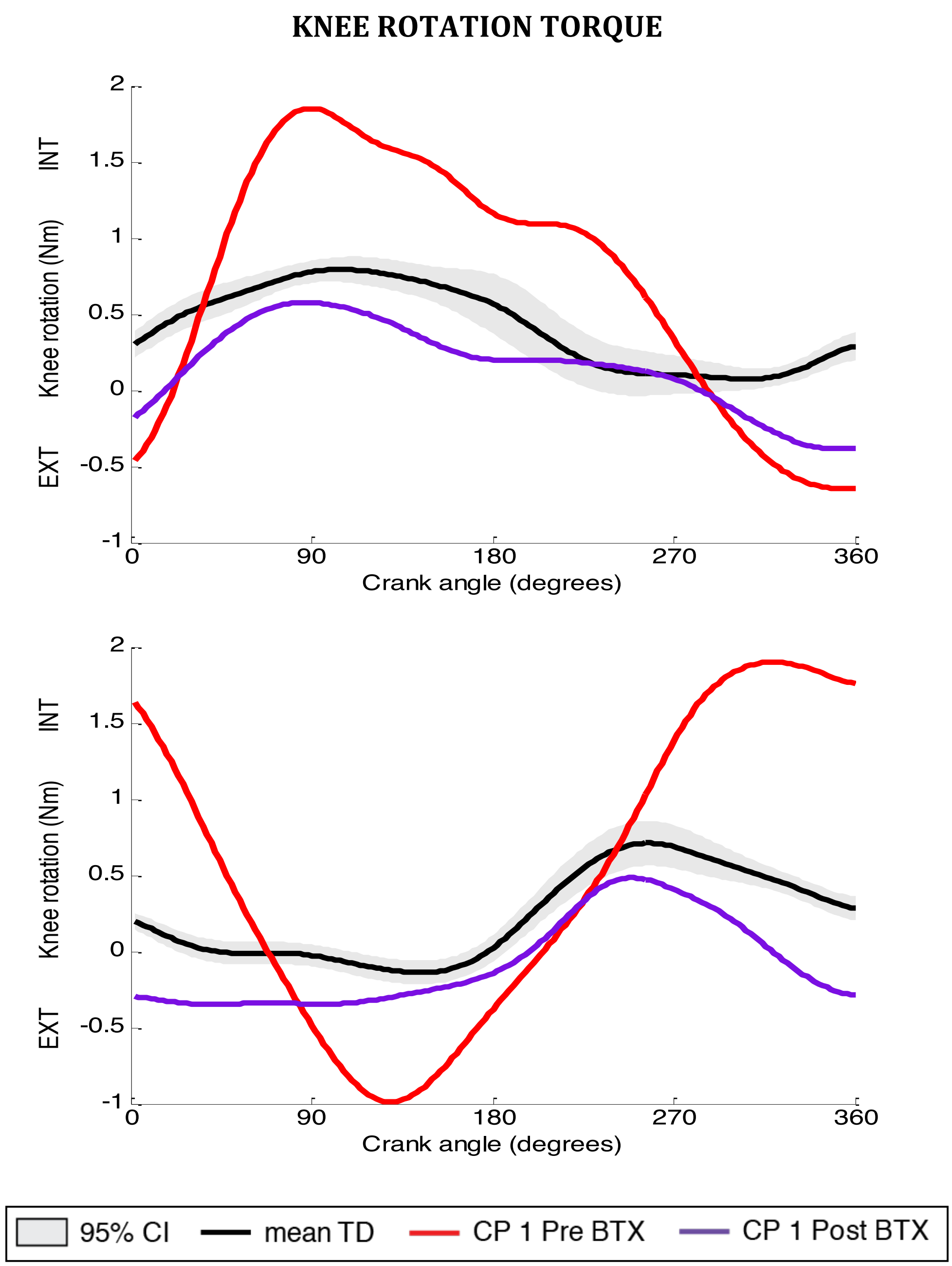


Figure A154: Knee rotation torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

ANKLE FLEXION TORQUE

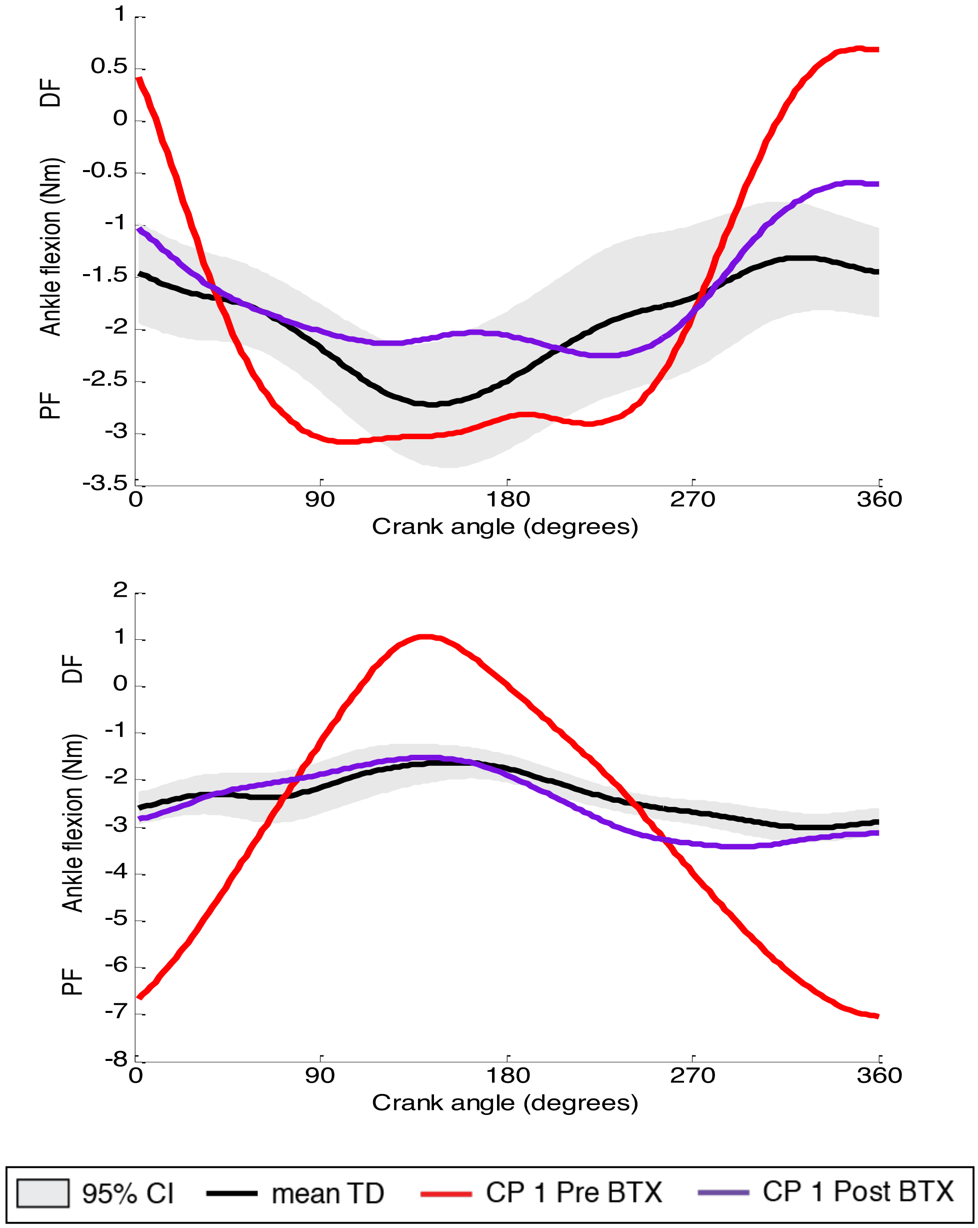


Figure A155: Ankle flexion torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

SUBTALAR ROTATION TORQUE

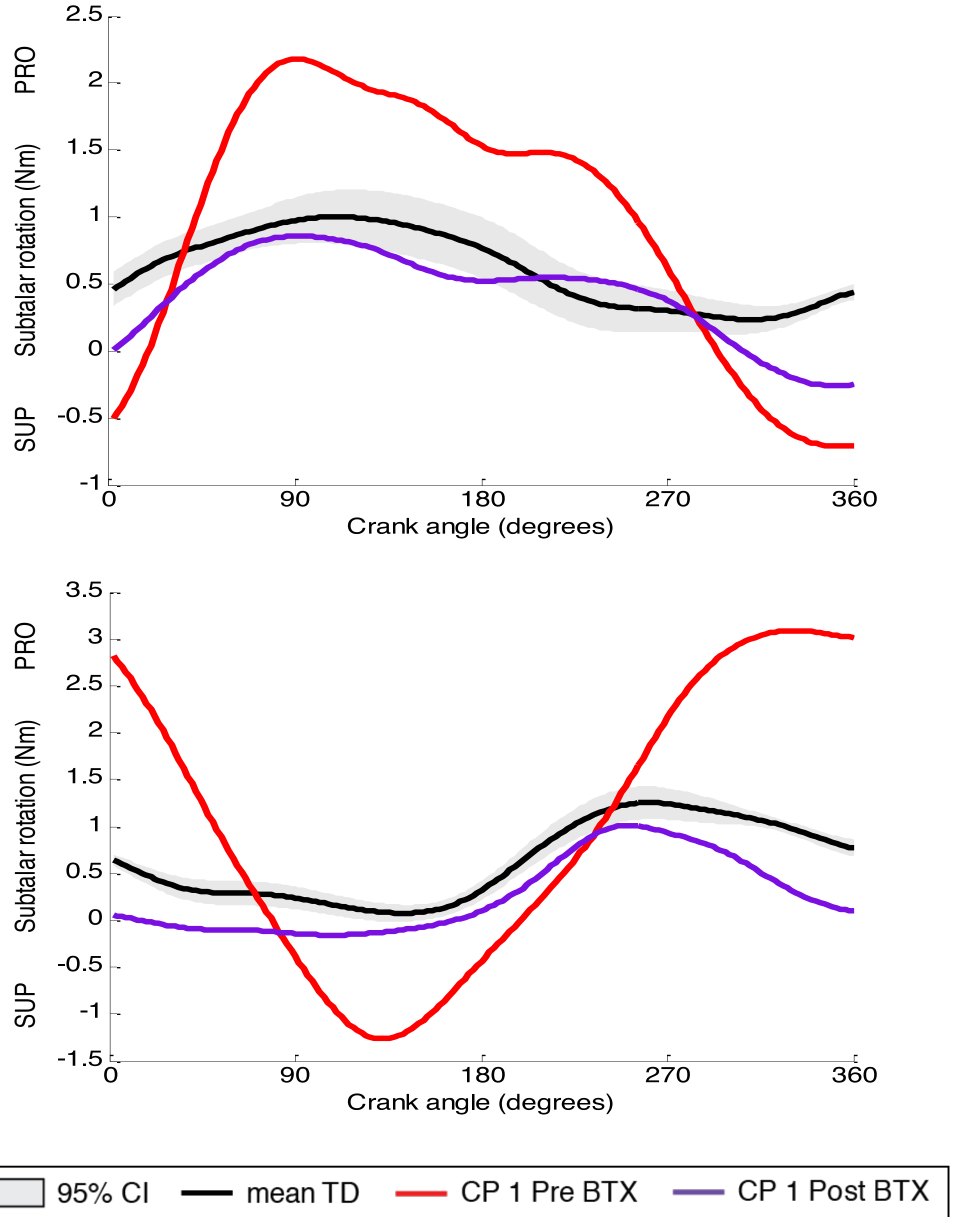


Figure A156: Subtalar rotation torques pre and post BTX at 40 RPM at treated (top) and non-treated (bottom) leg.

Appendix AI: Joint torques participant 1 at 50 RPM in Study 3

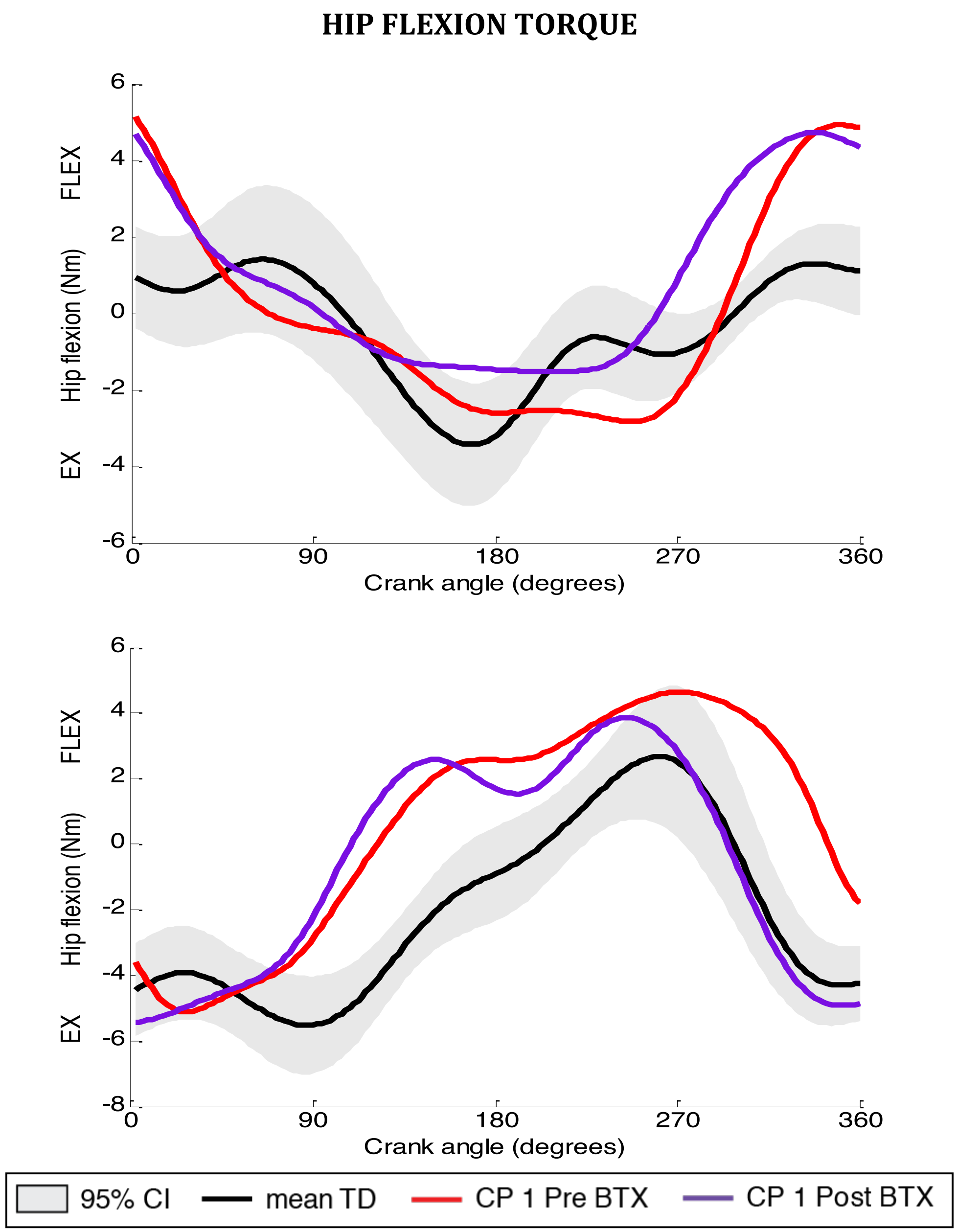


Figure A157: Hip flexion torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

HIP ADDUCTION TORQUE

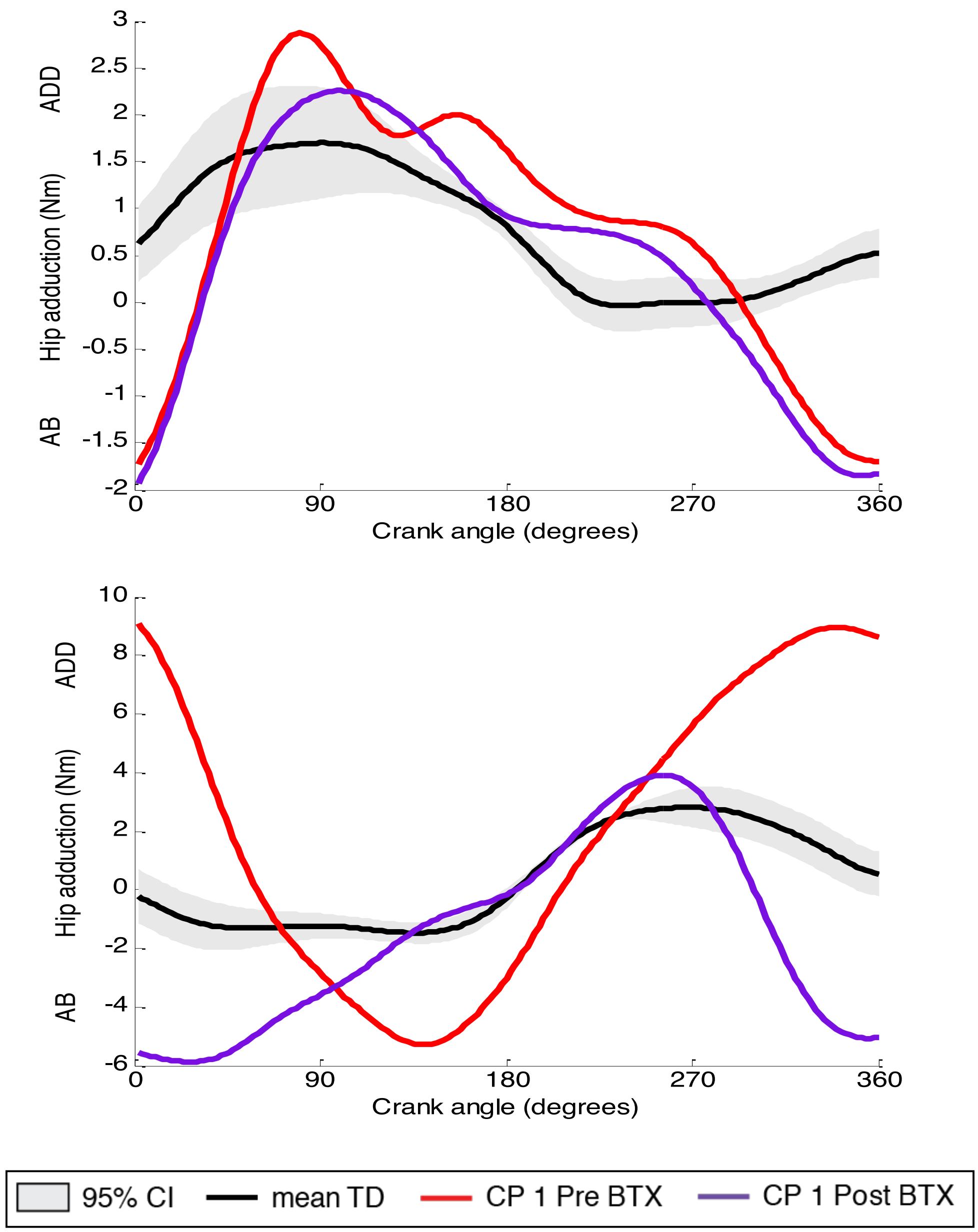


Figure A158: Hip adduction torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

HIP ROTATION TORQUE

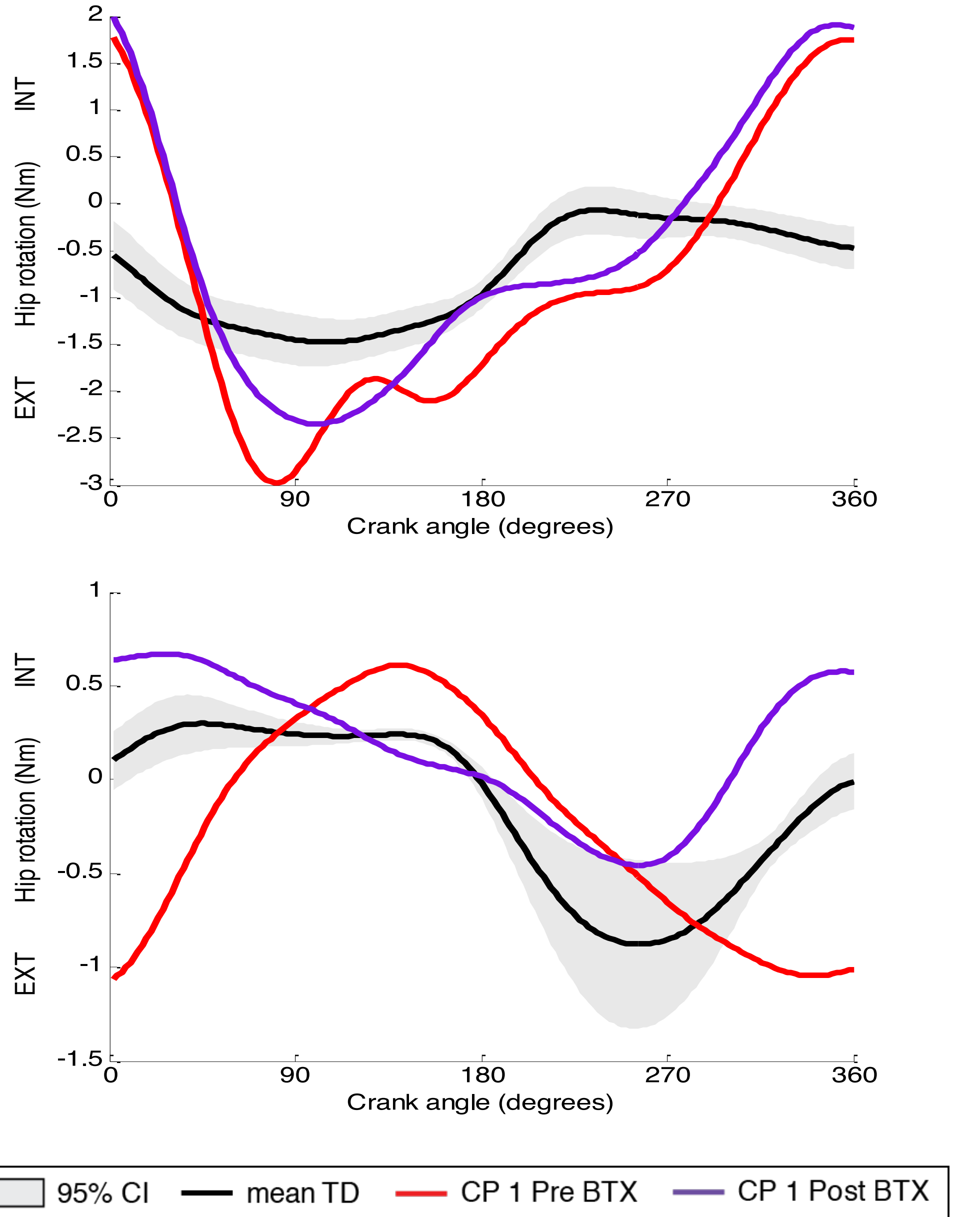


Figure A159: Hip rotation torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

KNEE FLEXION TORQUE

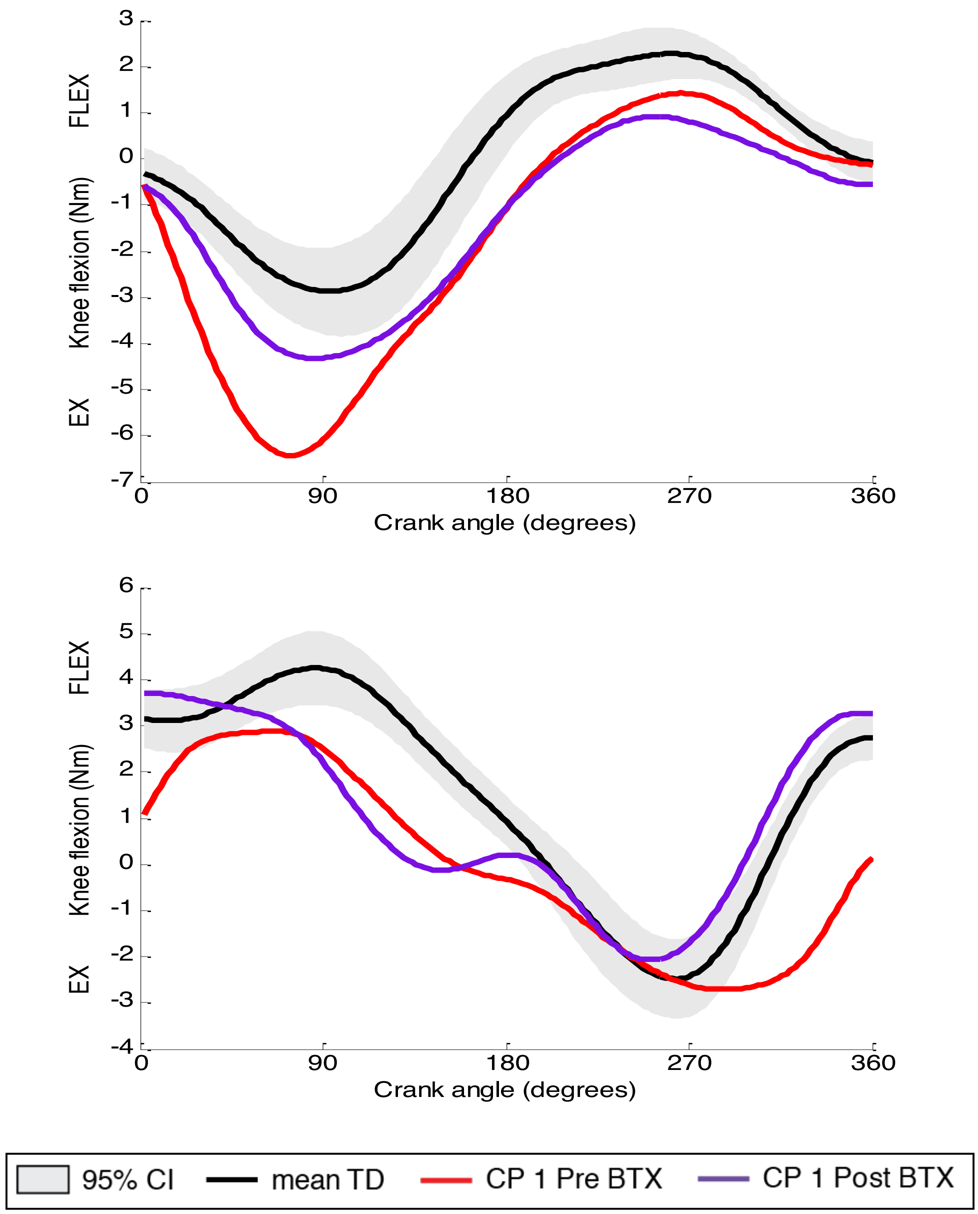


Figure A160: Knee flexion torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

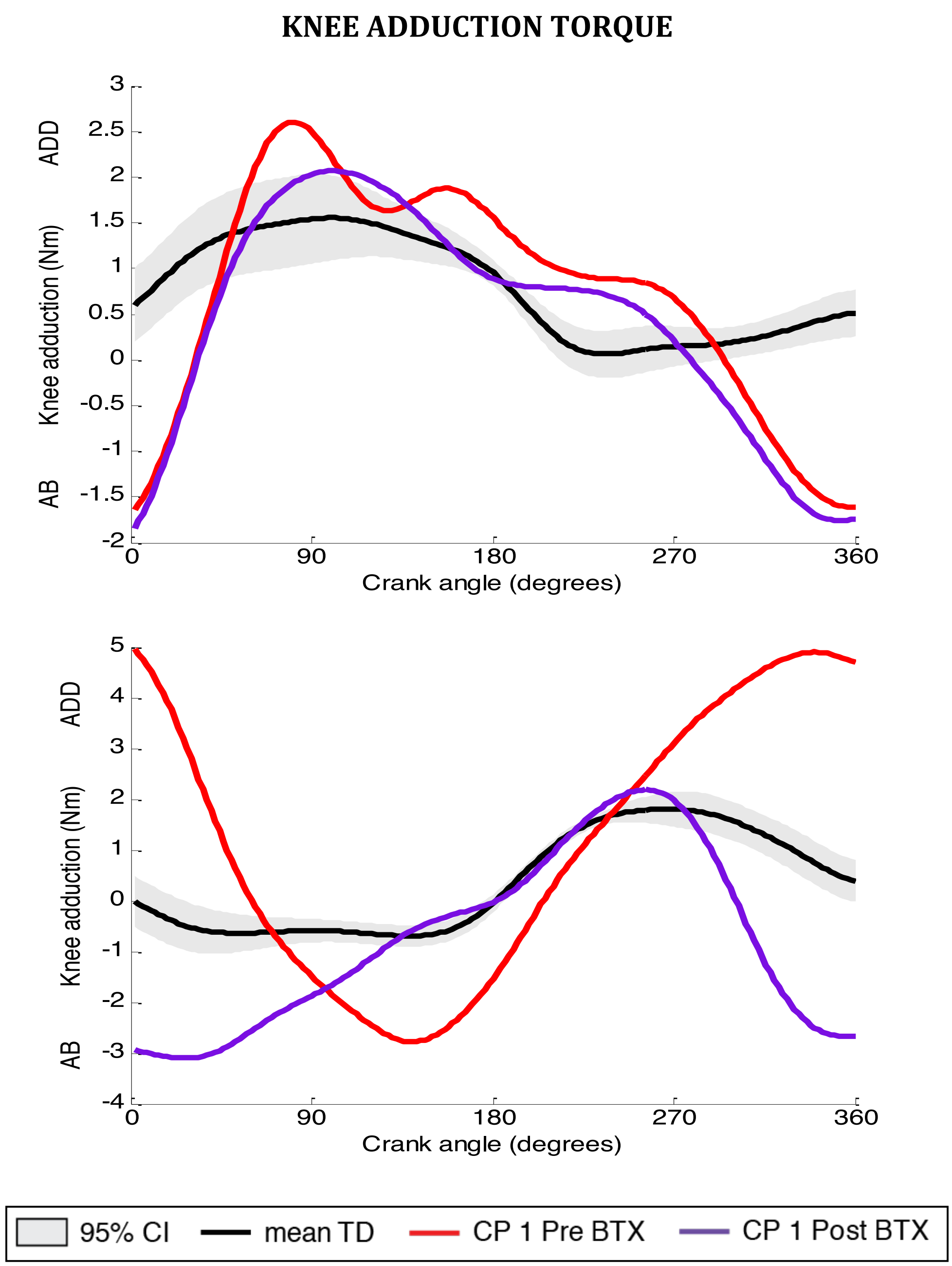


Figure A161: Knee adduction torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

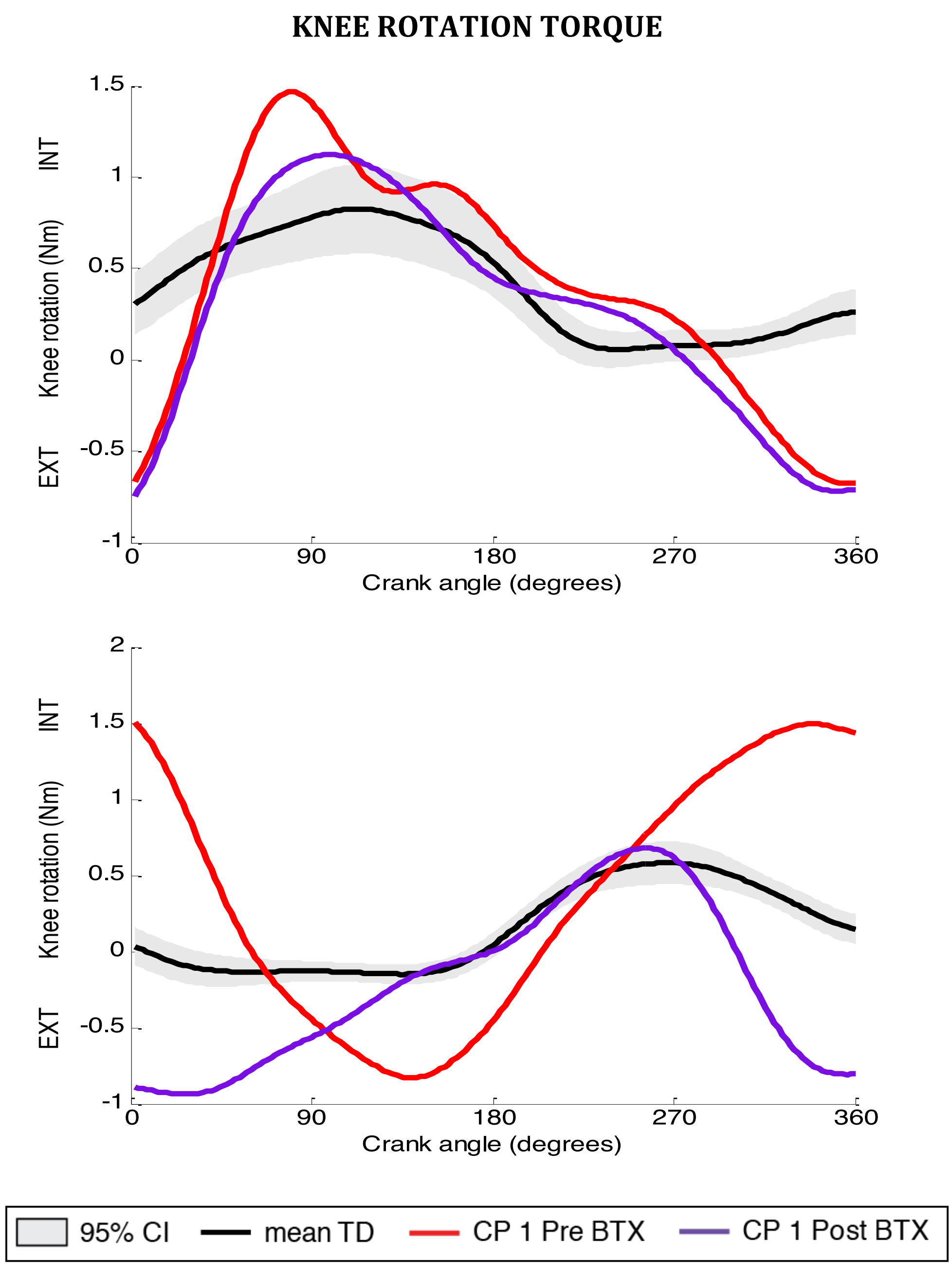


Figure A162: Knee rotation torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

ANKLE FLEXION TORQUE

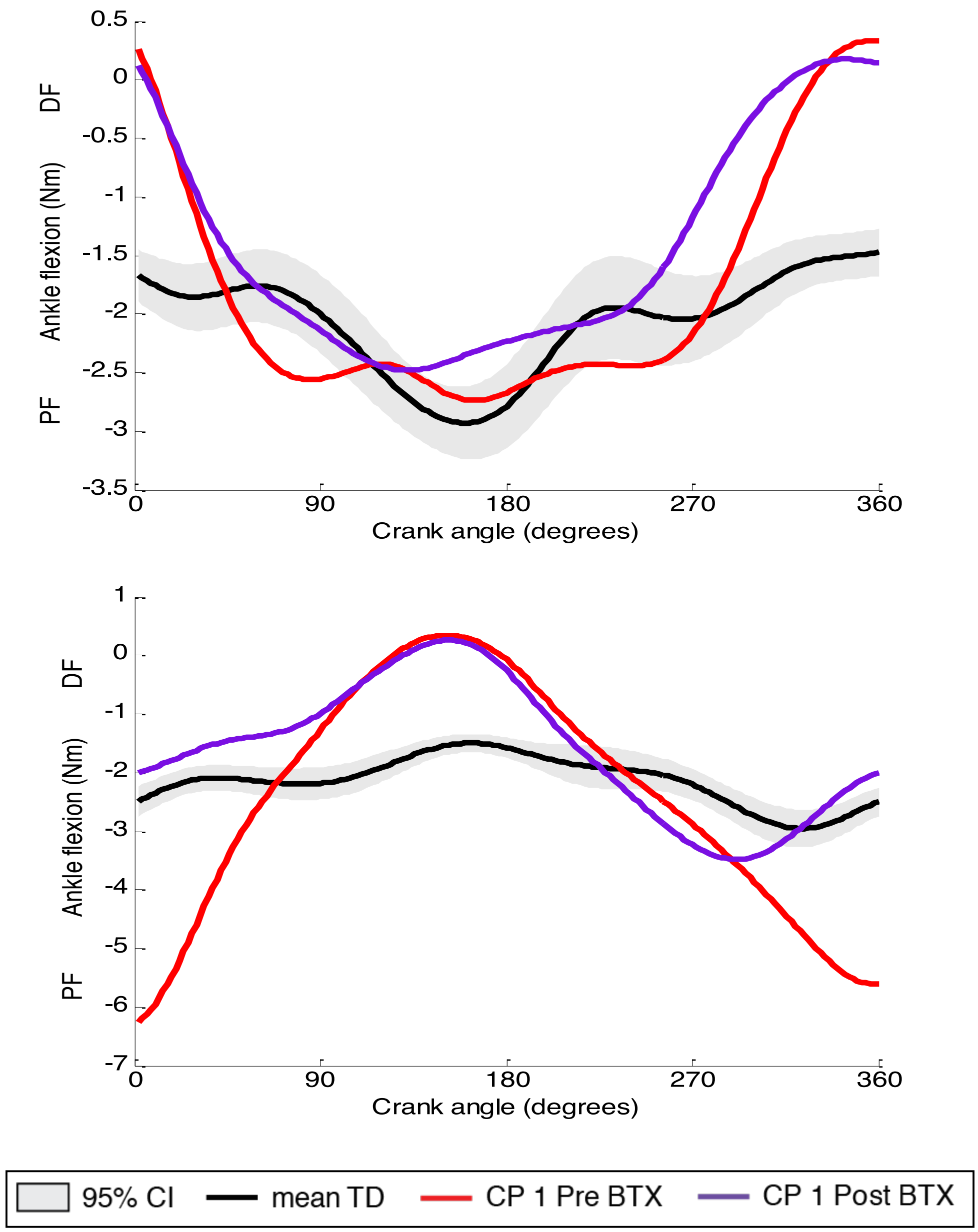


Figure A163: Ankle flexion torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

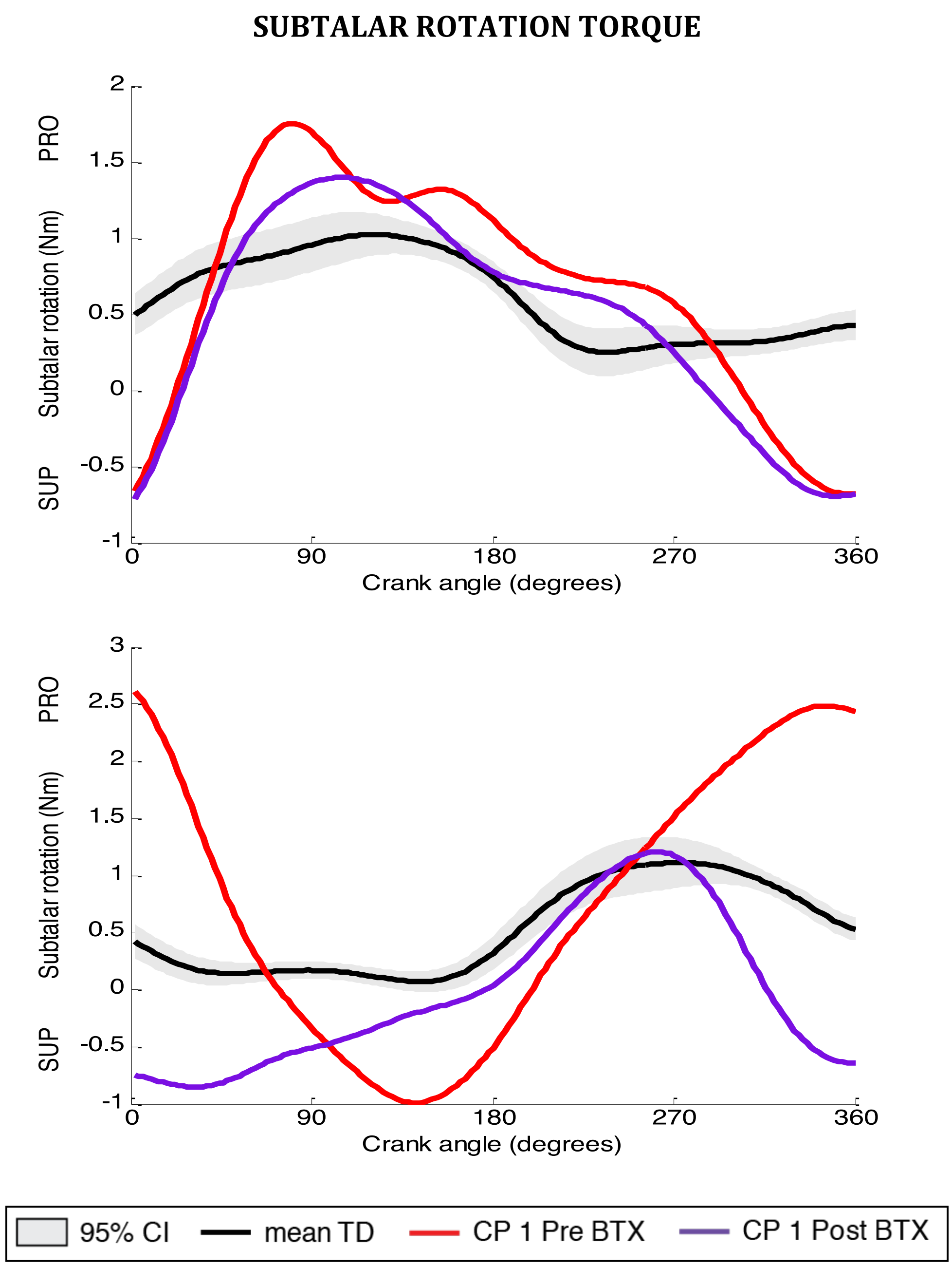


Figure A164: Subtalar rotation torques pre and post BTX at 50 RPM at treated (top) and non-treated (bottom) leg.

Appendix AJ: Joint torques participant 2 at 60 RPM in Study 3

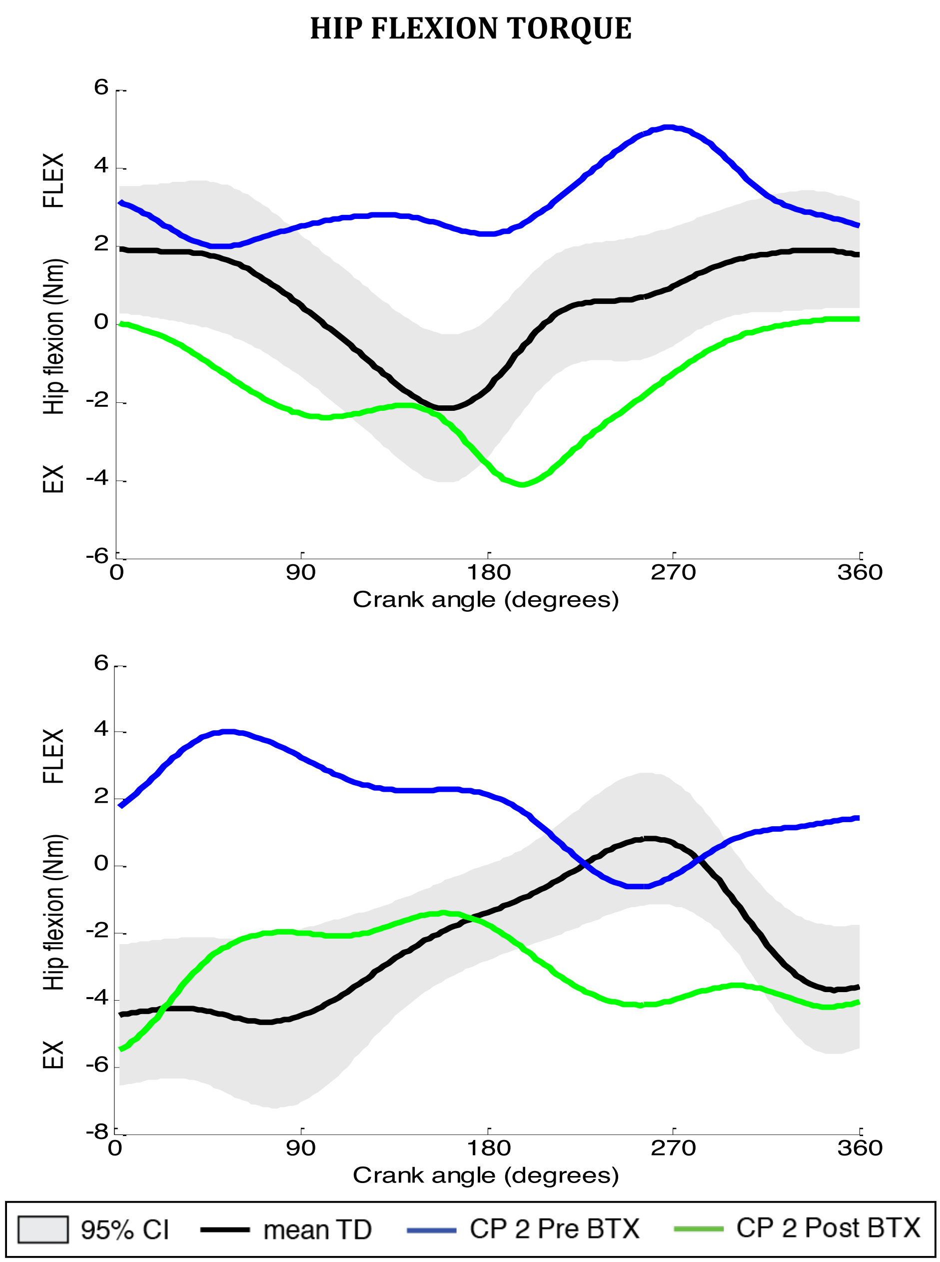


Figure A165: Hip flexion torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

HIP ADDUCTION TORQUE

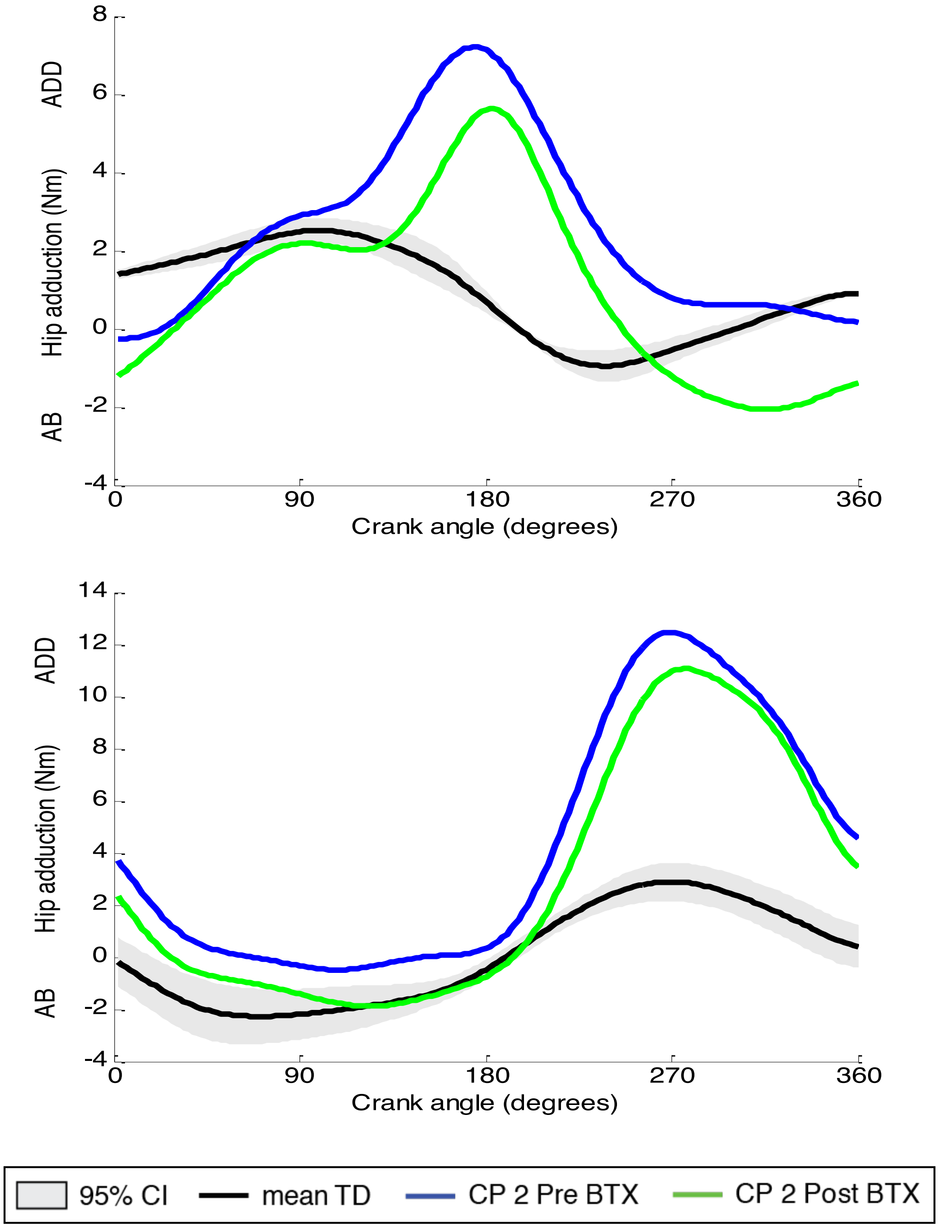


Figure A166: Hip adduction torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

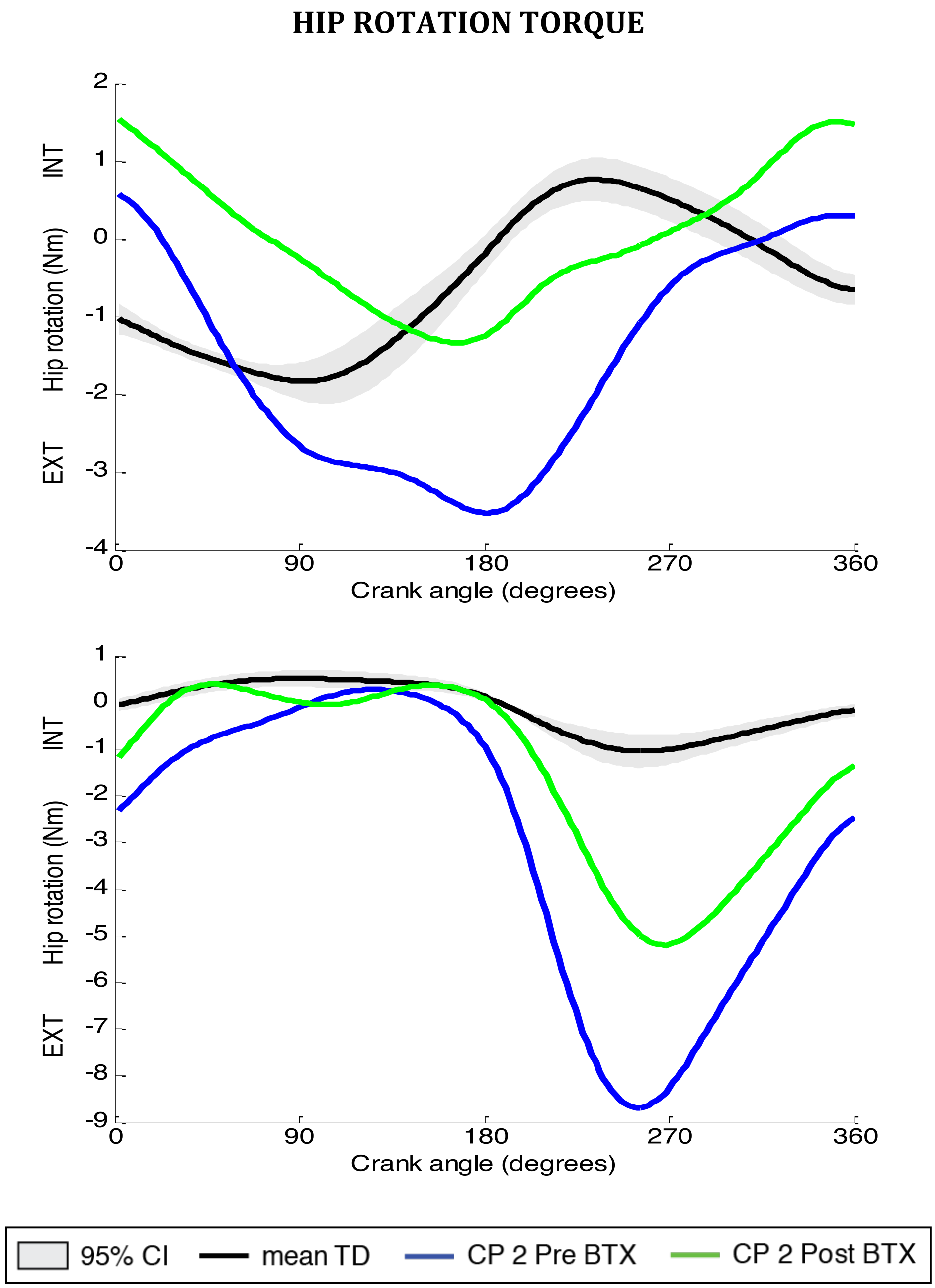


Figure A167: Hip rotation torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

KNEE FLEXION TORQUE

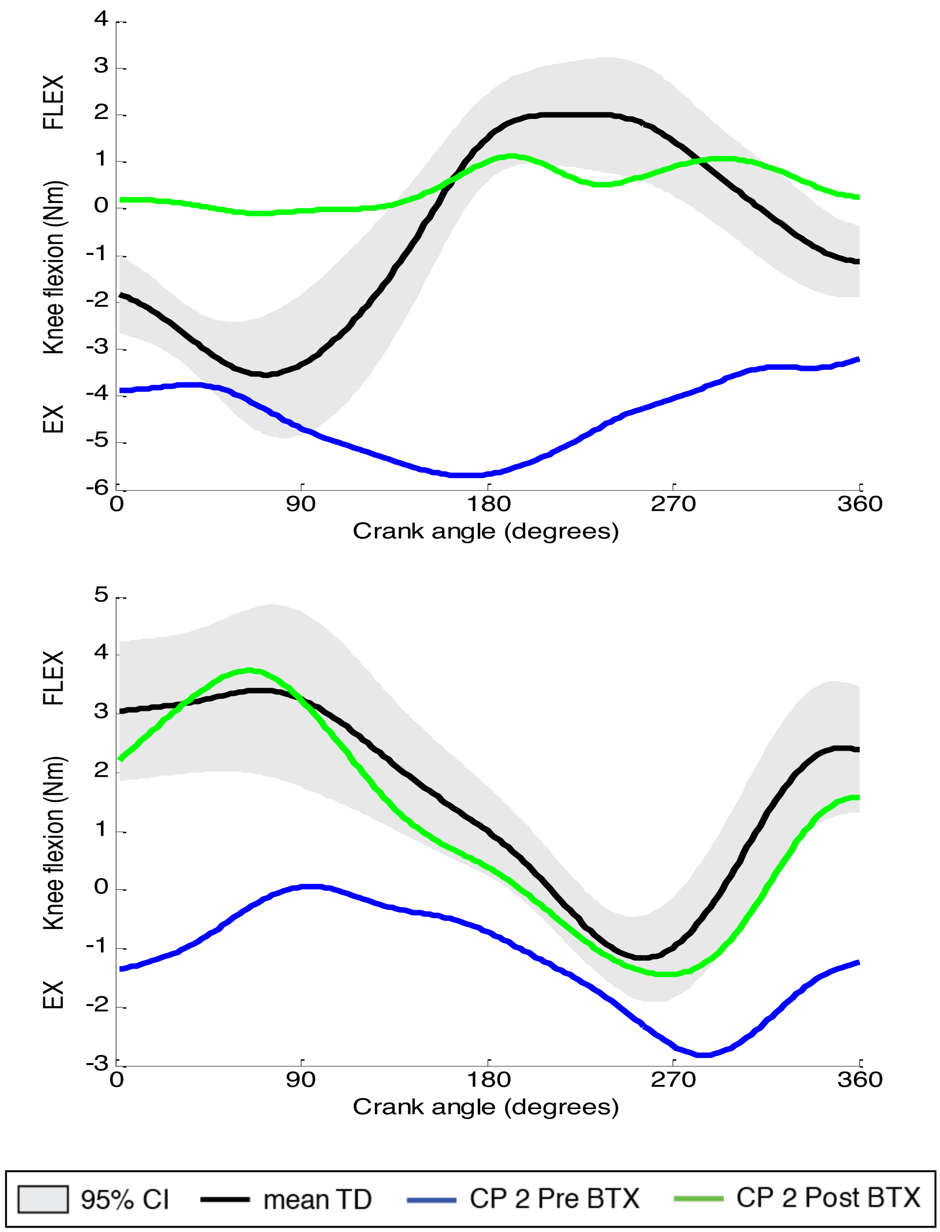


Figure A168: Knee flexion torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

KNEE ADDUCTION TORQUE

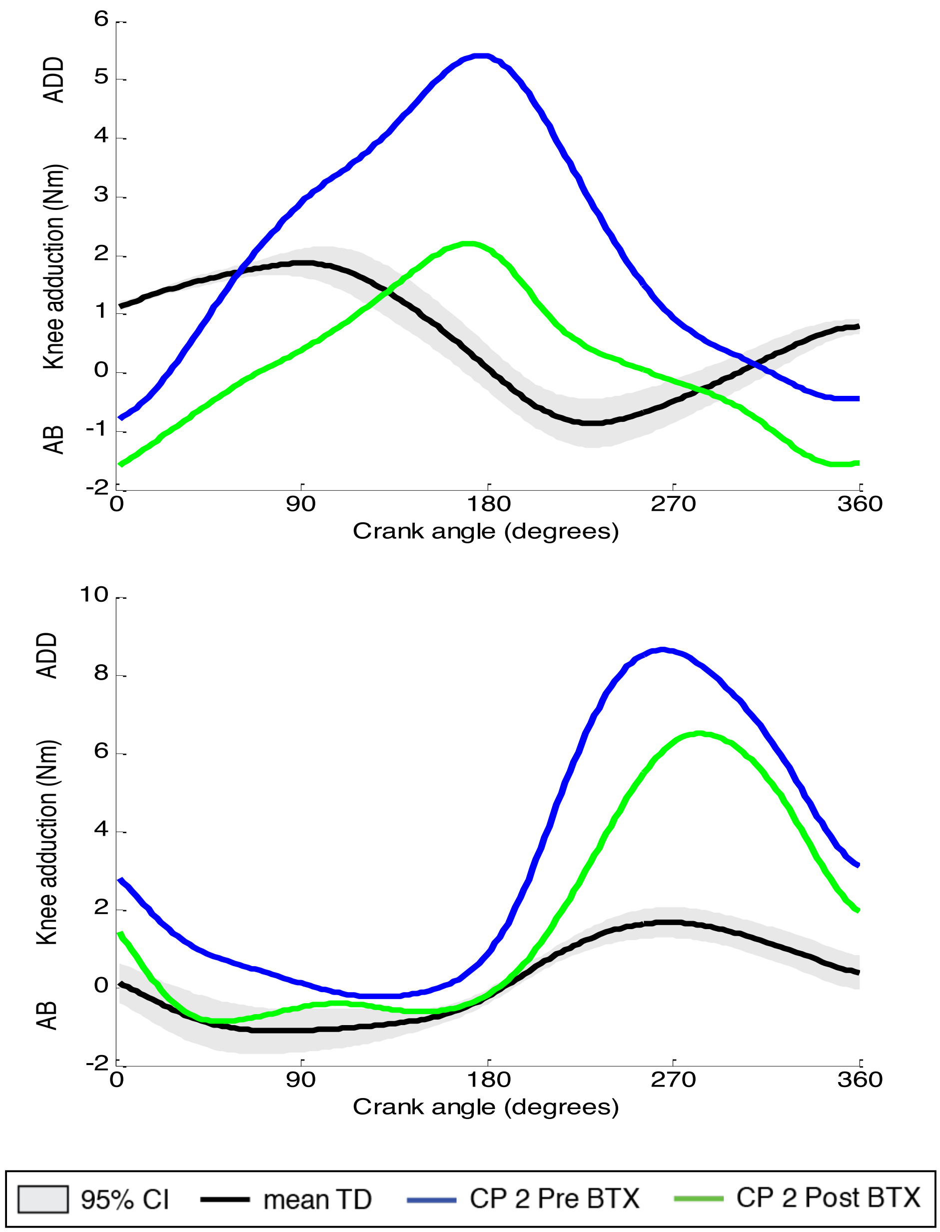


Figure A169: Knee adduction torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

KNEE ROTATION TORQUE

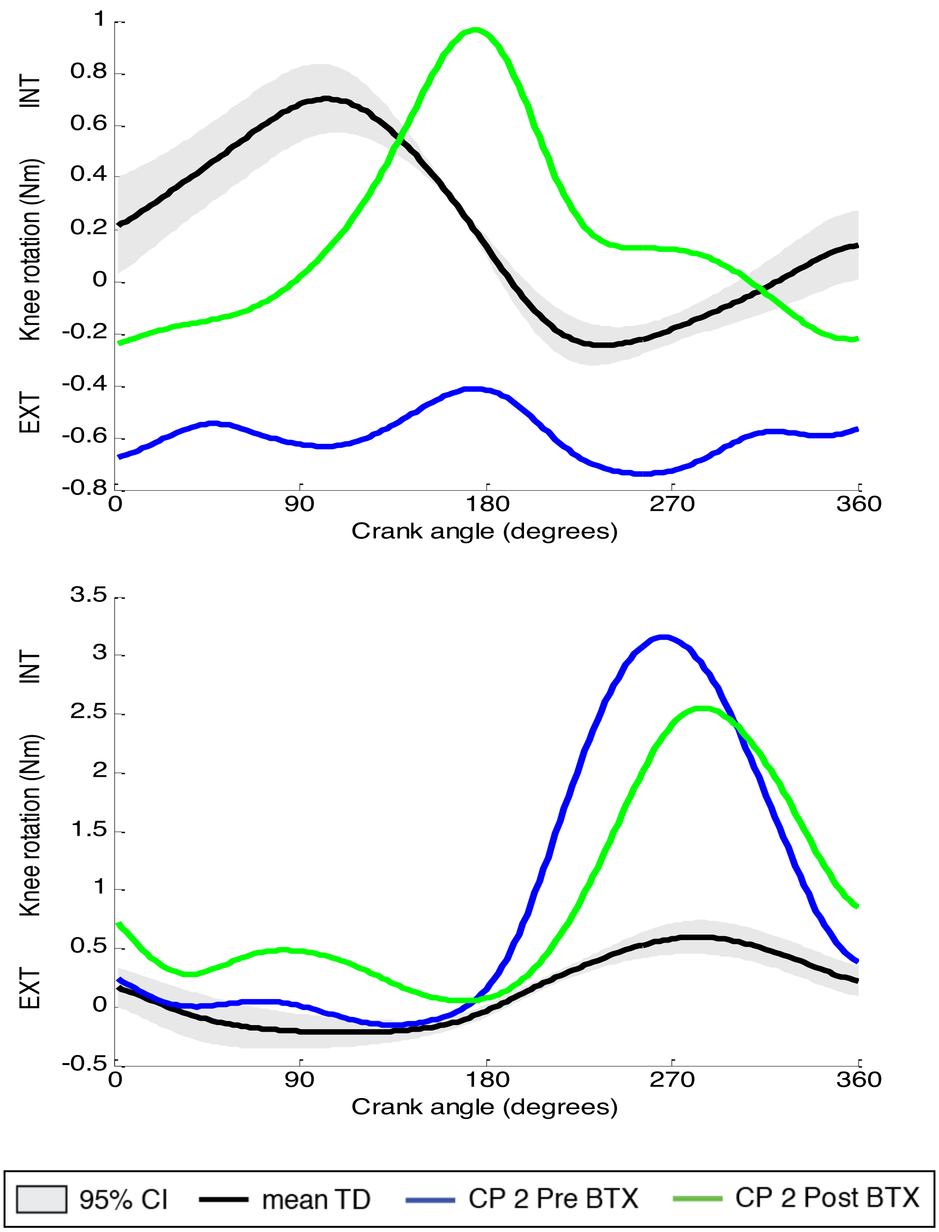


Figure A170: Knee rotation torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

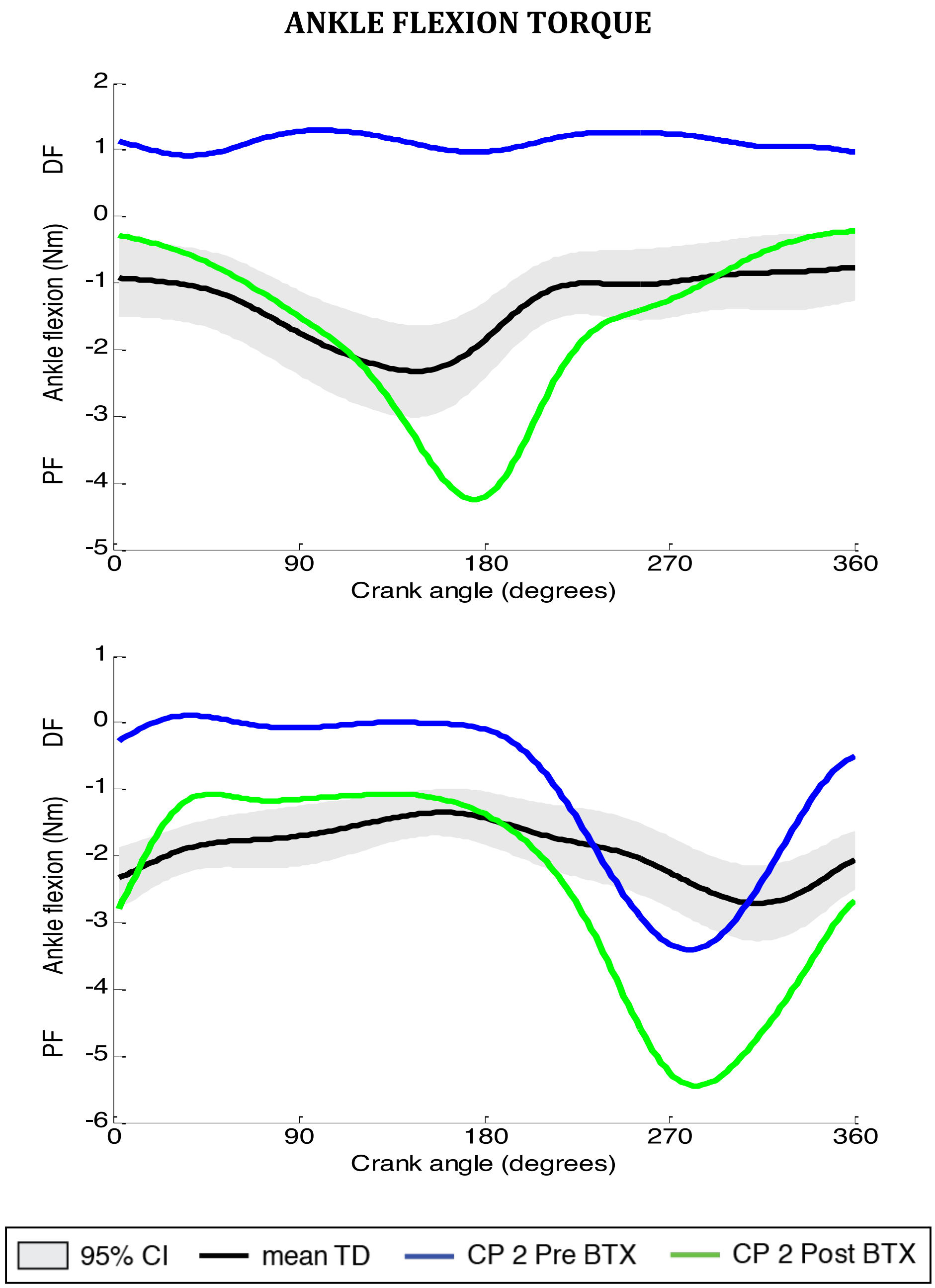


Figure A171: Ankle flexion torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

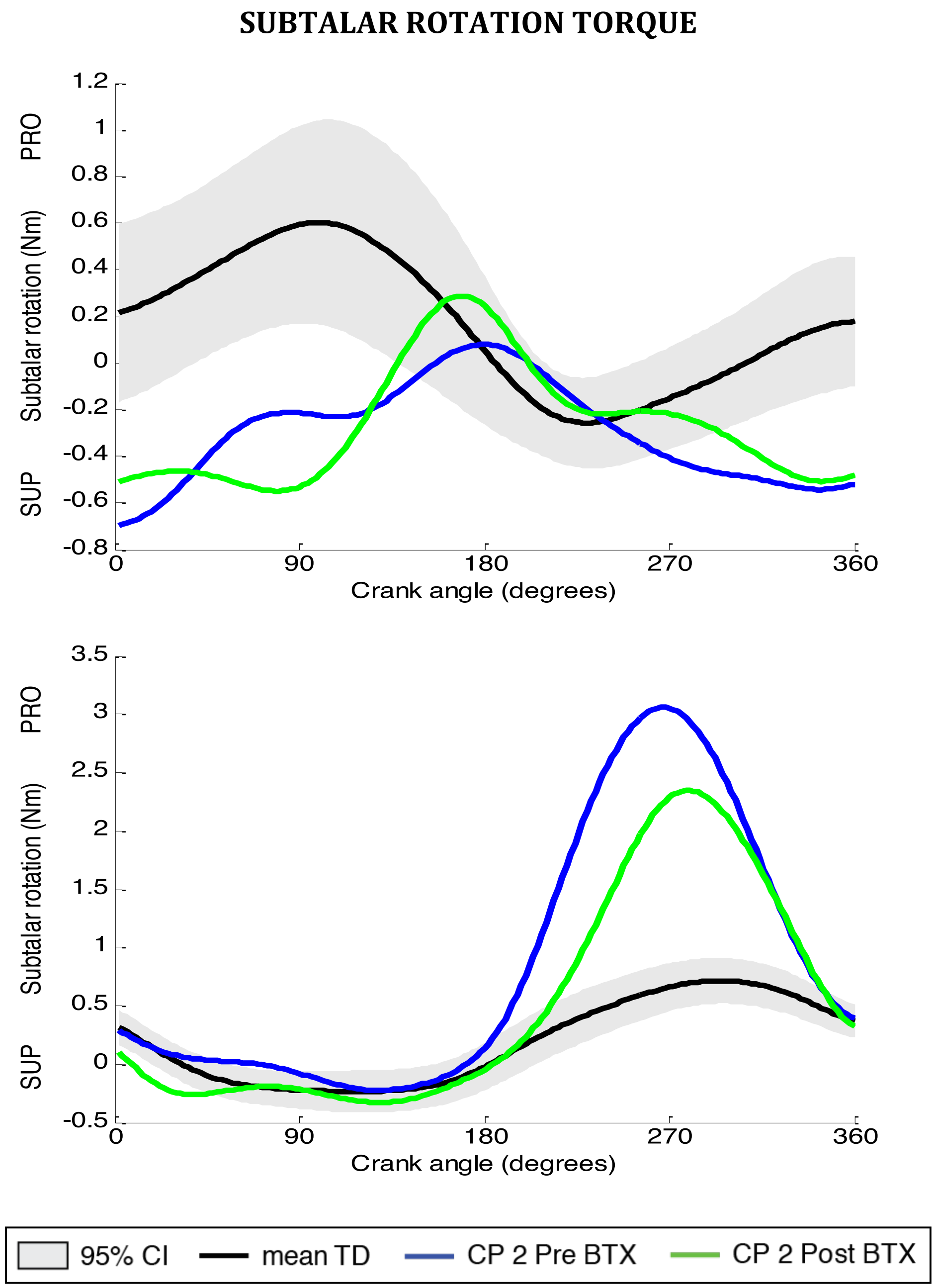


Figure A172: Subtalar rotation torques pre and post BTX at 60 RPM at treated (top) and non-treated (bottom) leg.

APPENDIX AK: JOINT TORQUES PARTICIPANT 2 AT 70 RPM IN STUDY 3

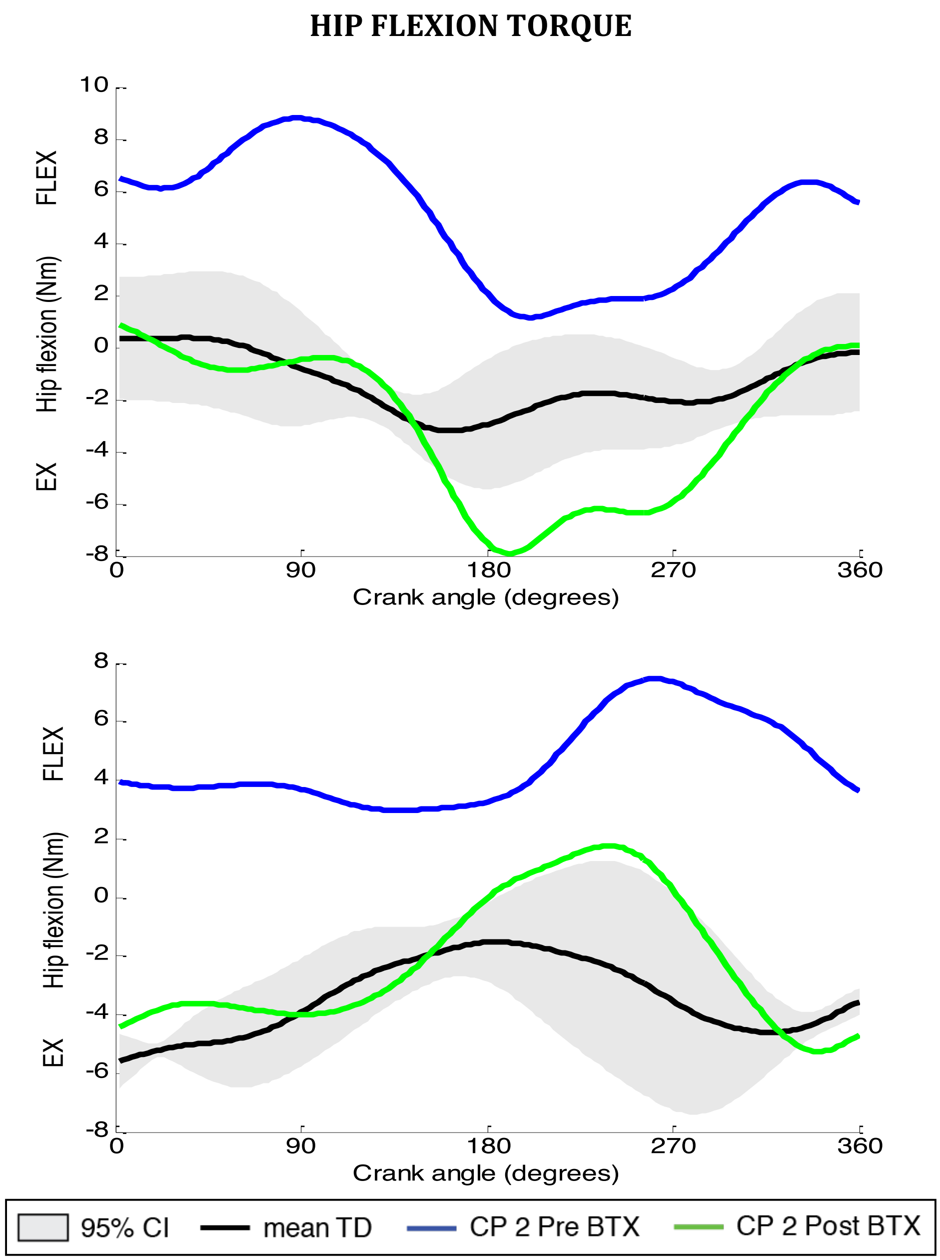


Figure A173: Hip flexion torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

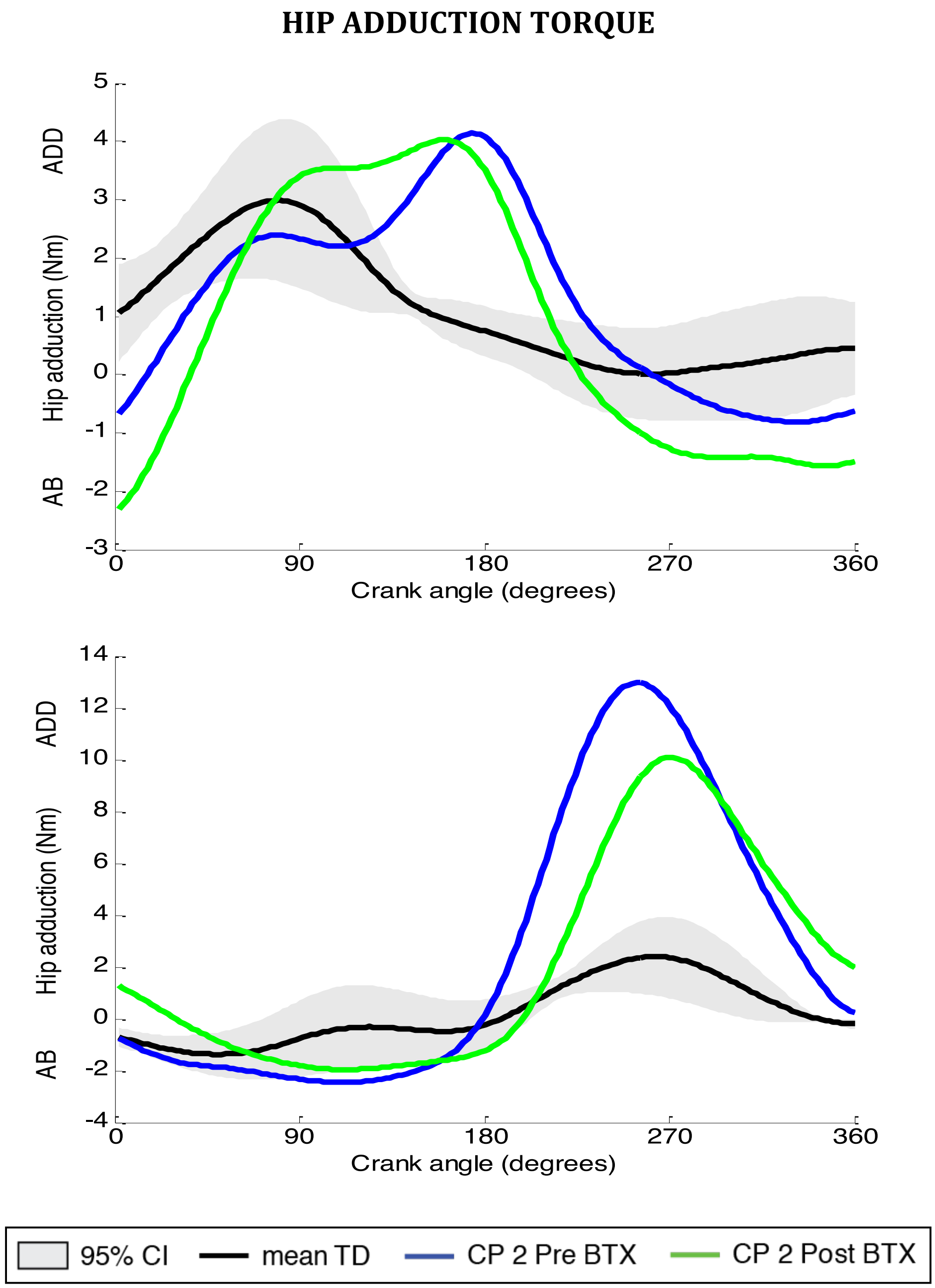


Figure A174: Hip adduction torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

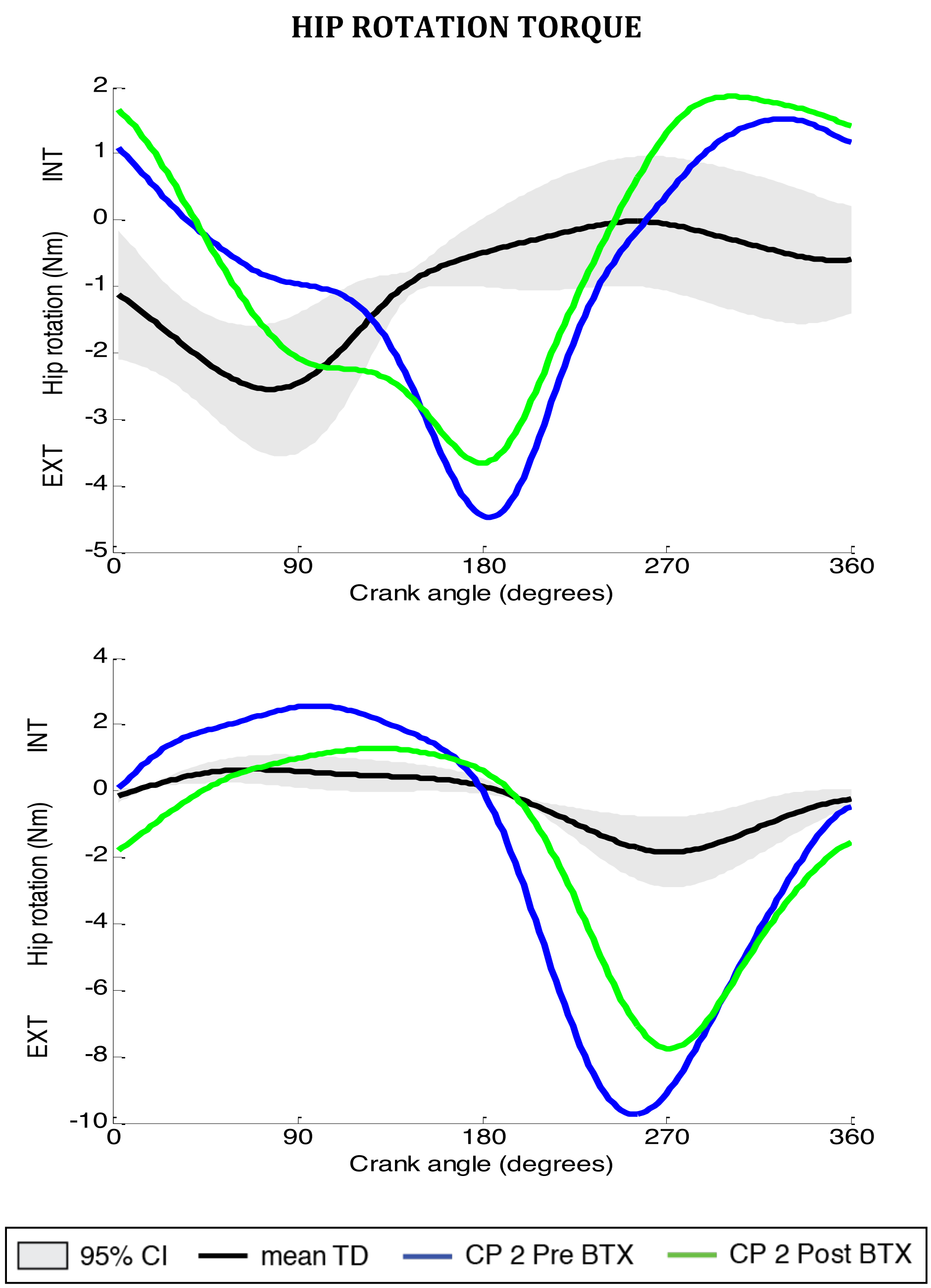


Figure A175: Hip rotation torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

KNEE FLEXION TORQUE

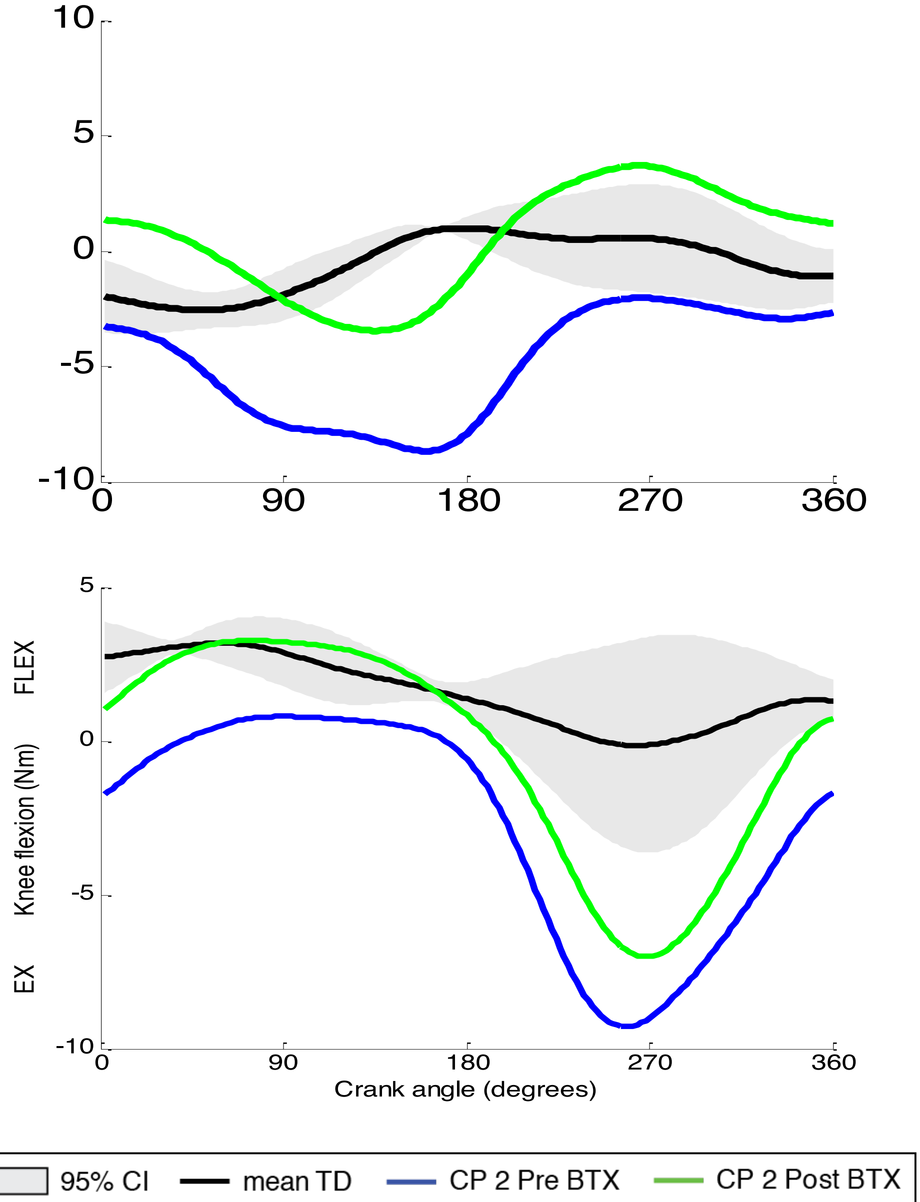


Figure A176: Knee flexion torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

KNEE ADDUCTION TORQUE

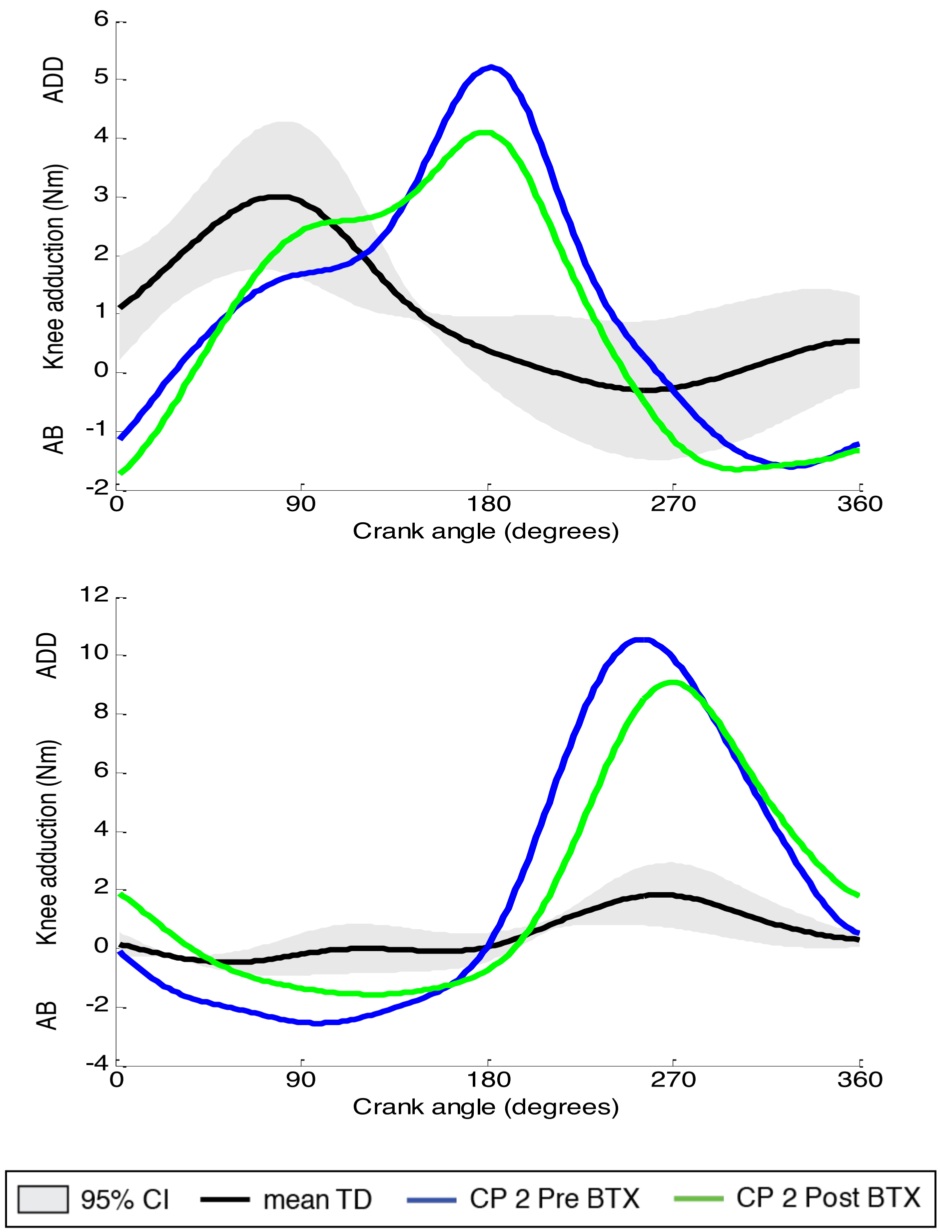


Figure A177: Knee adduction torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

KNEE ROTATION TORQUE

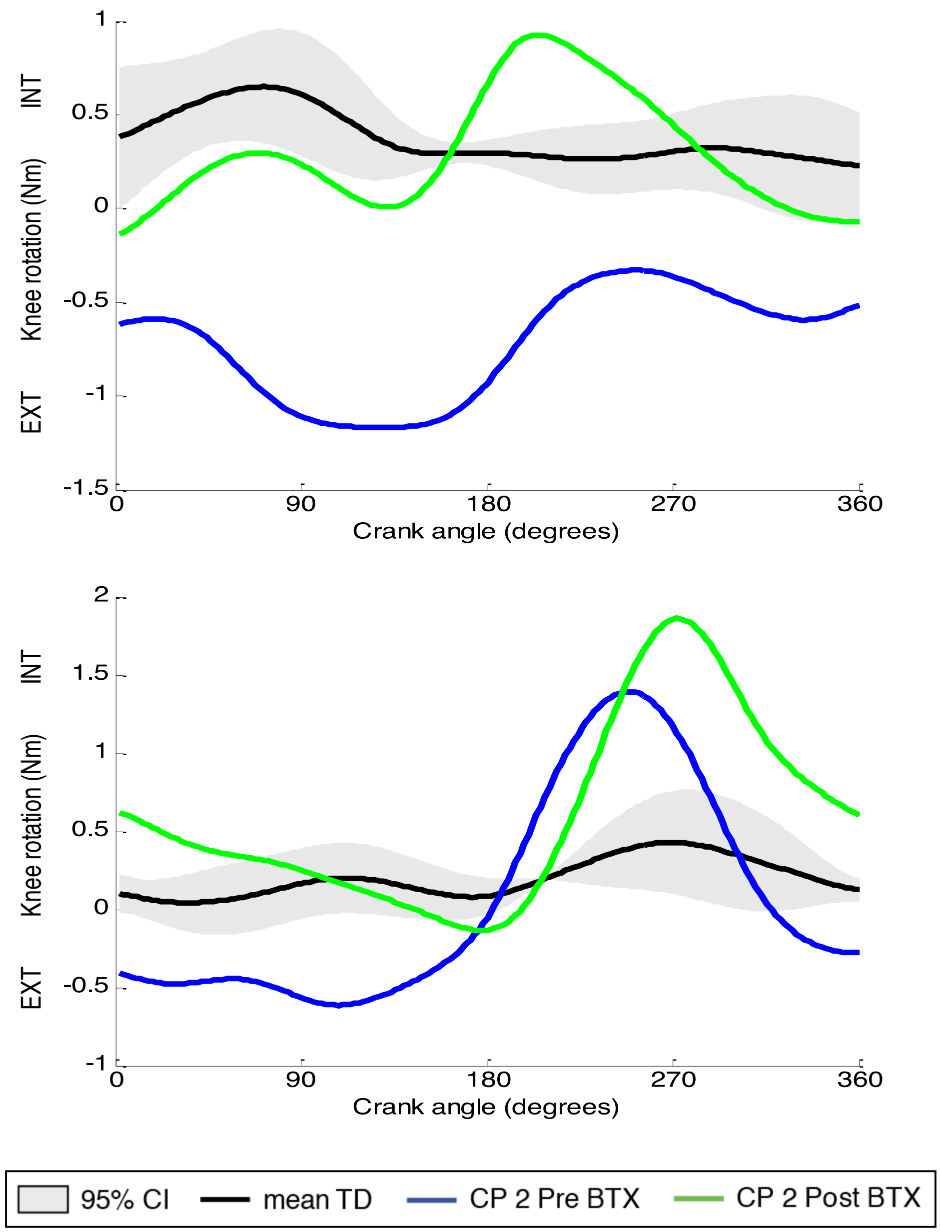


Figure A178: Knee rotation torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

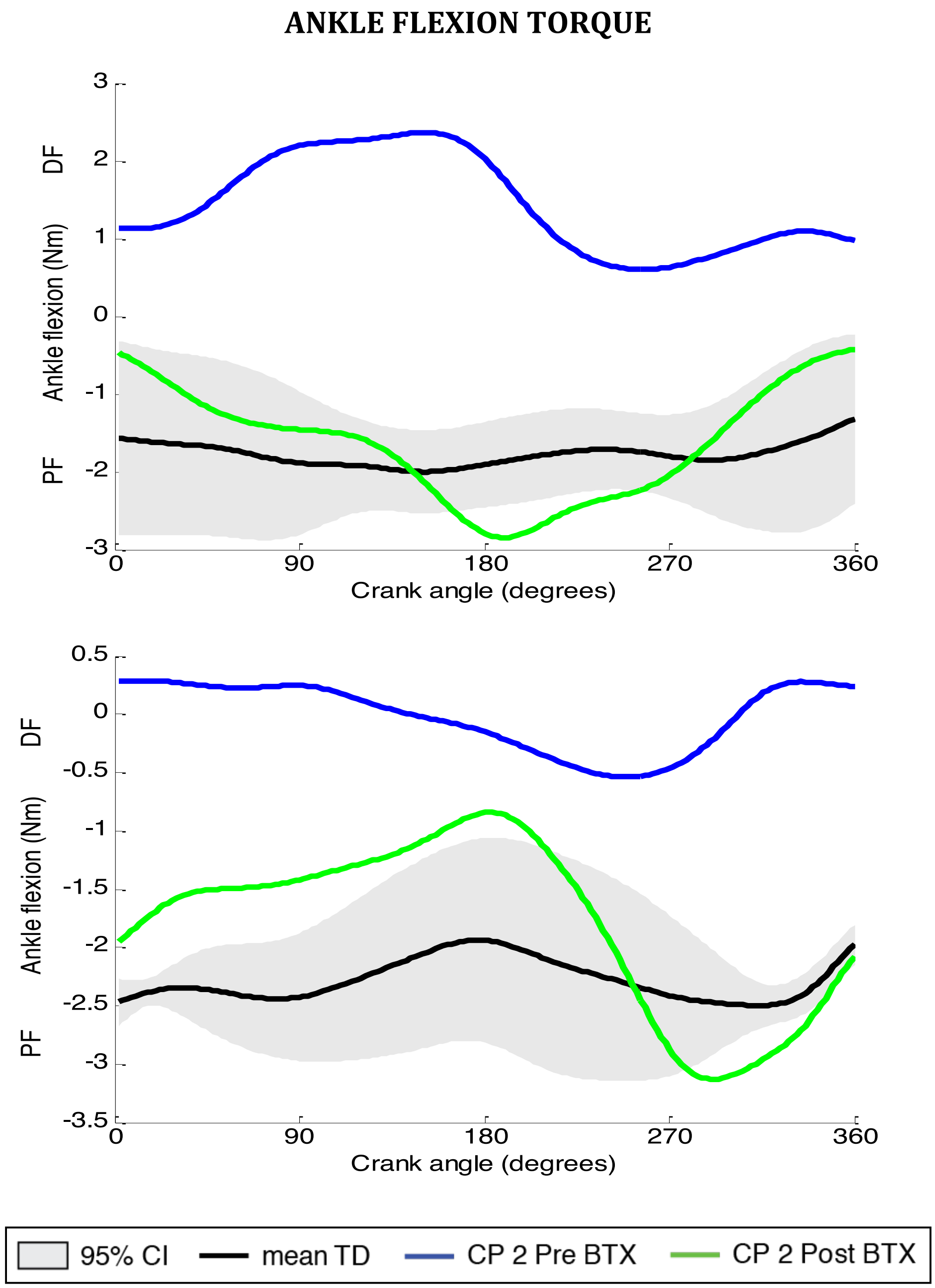


Figure A179: Ankle flexion torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

SUBTALAR ROTATION TORQUE

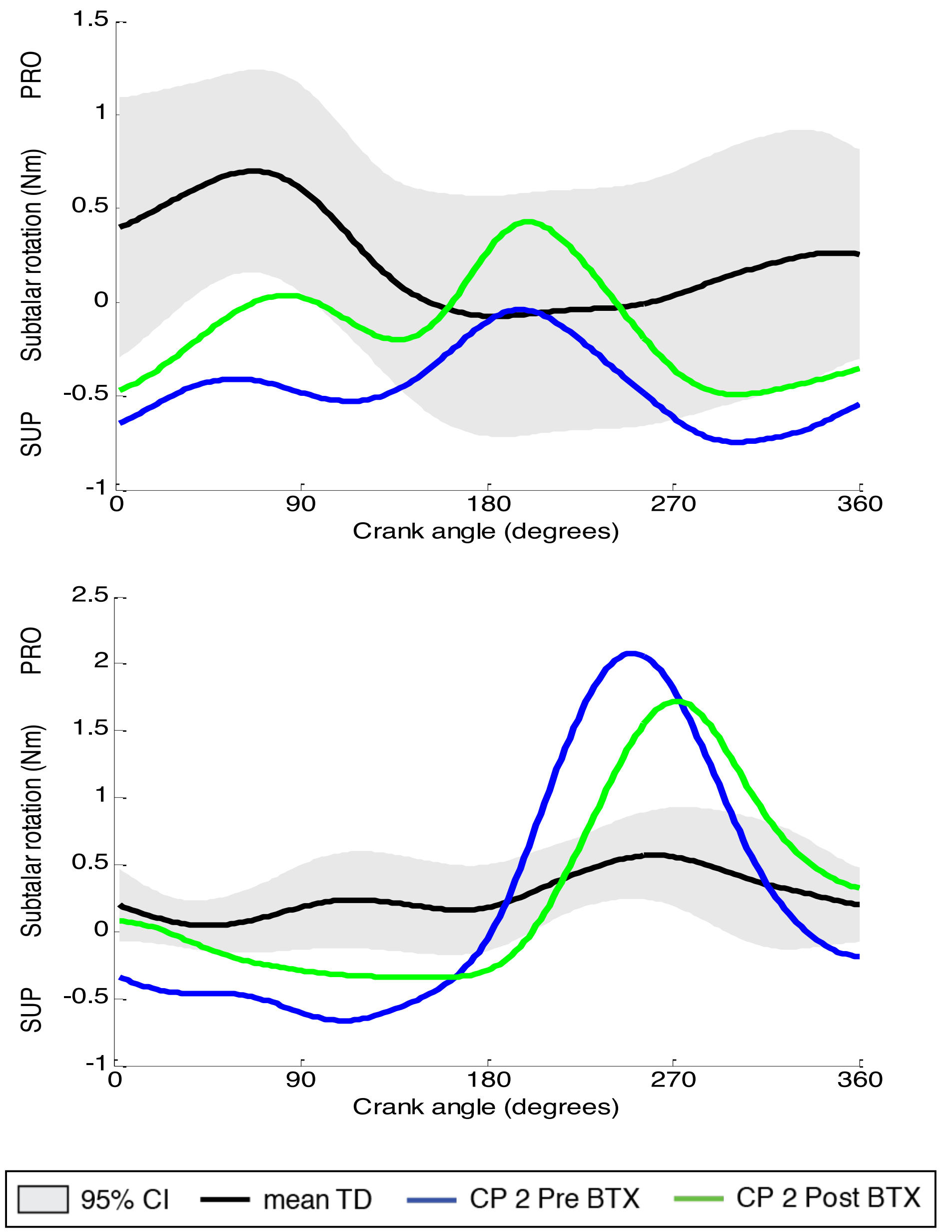


Figure A180: Subtalar rotation torques pre and post BTX at 70 RPM at treated (top) and non-treated (bottom) leg.

APPENDIX AL: JOINT POWERS PER CADENCE IN STUDY 3

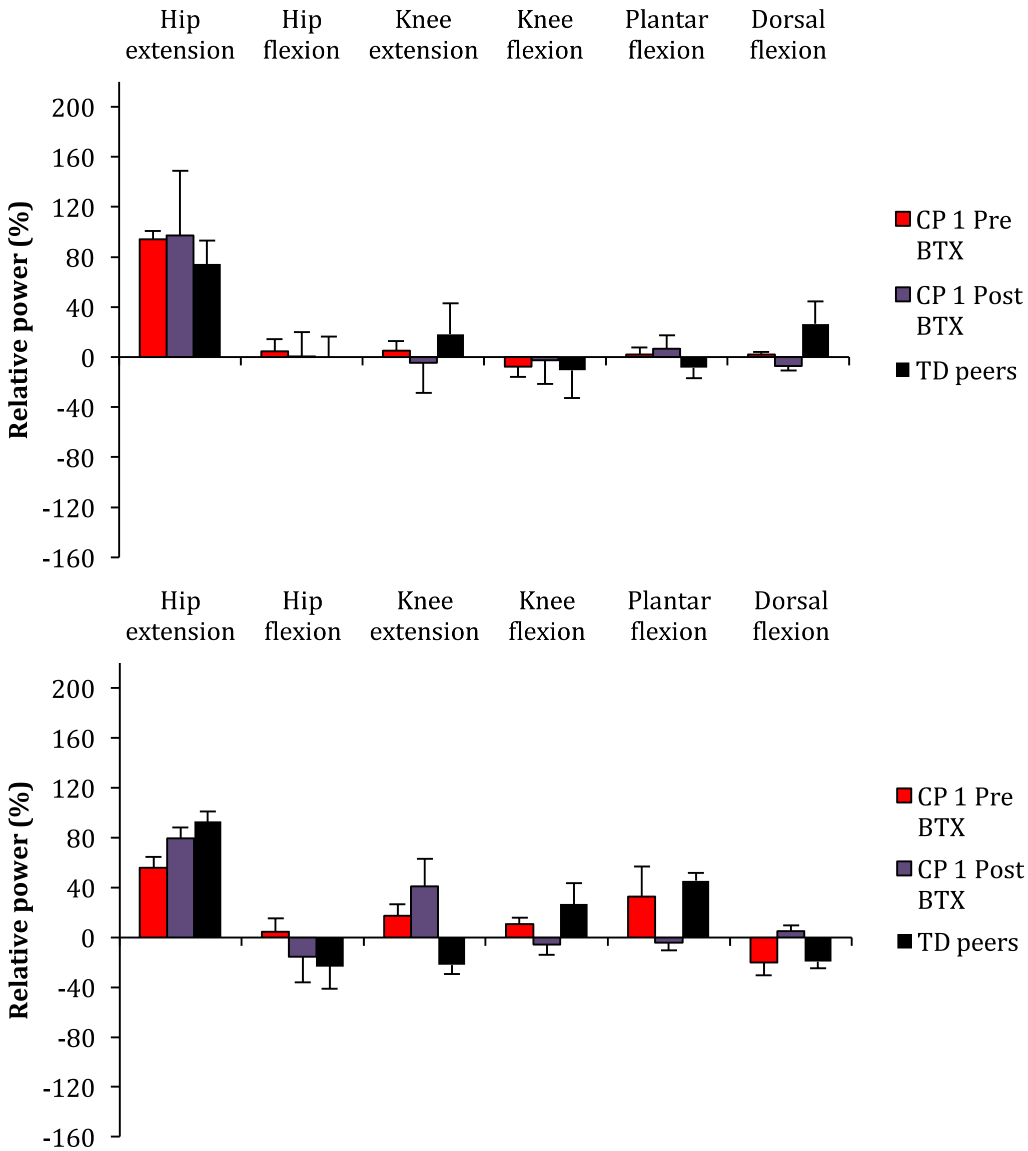


Figure A181: Joint powers produced at 30 RPM by treated (top) and non-treated (bottom) leg.

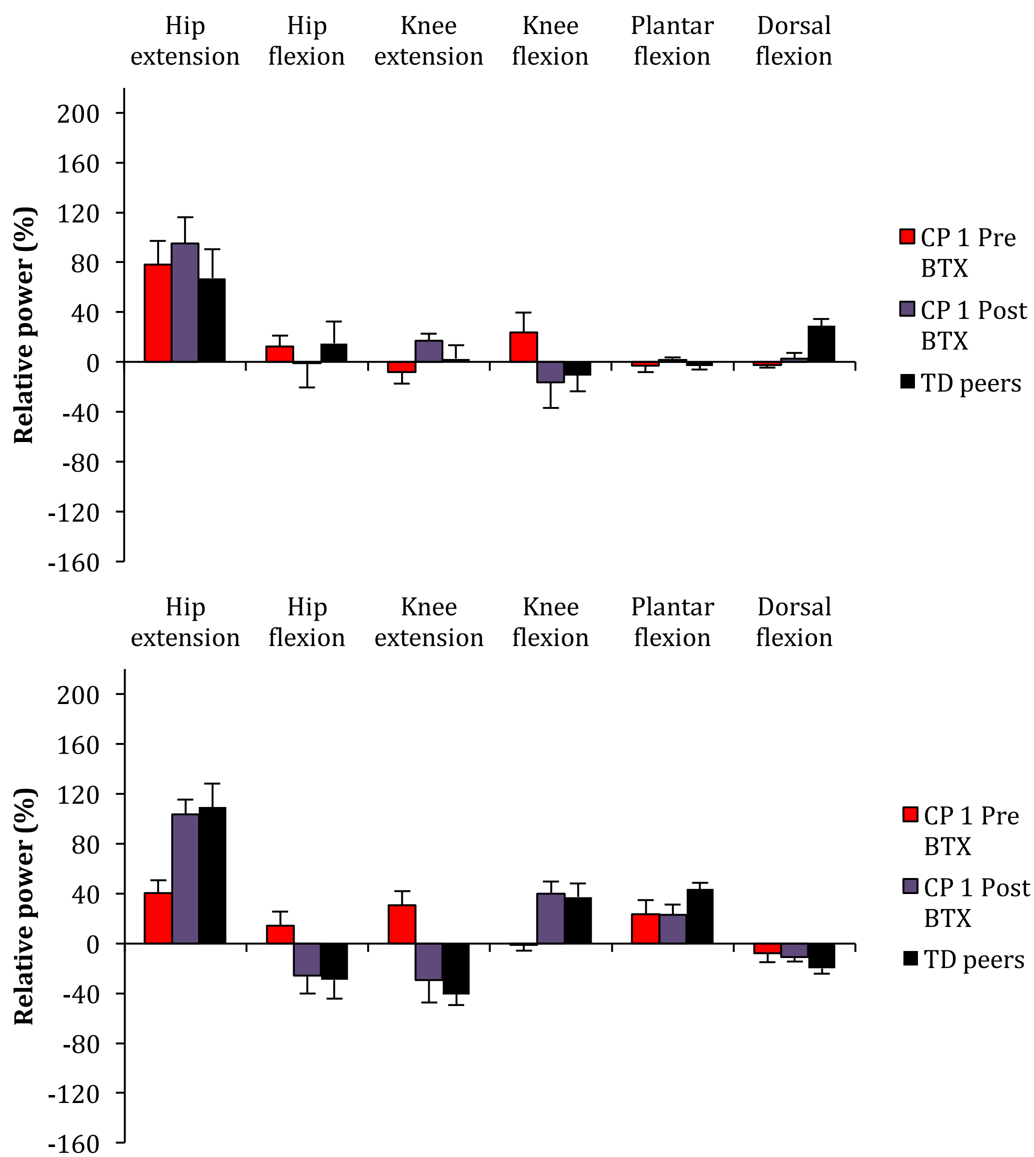
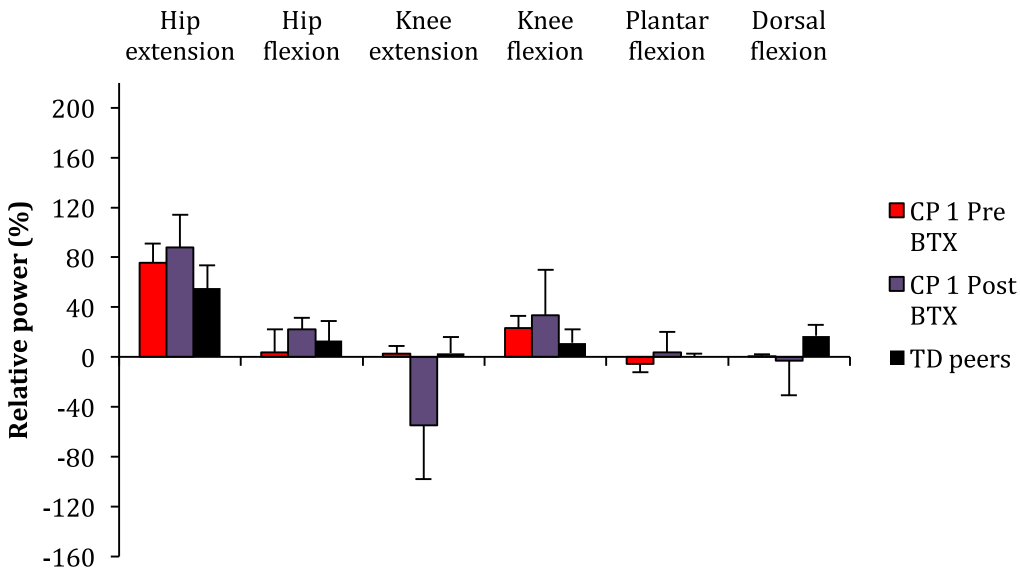


Figure A182: Joint powers produced at 40 RPM by treated (top) and non-treated (bottom) leg.



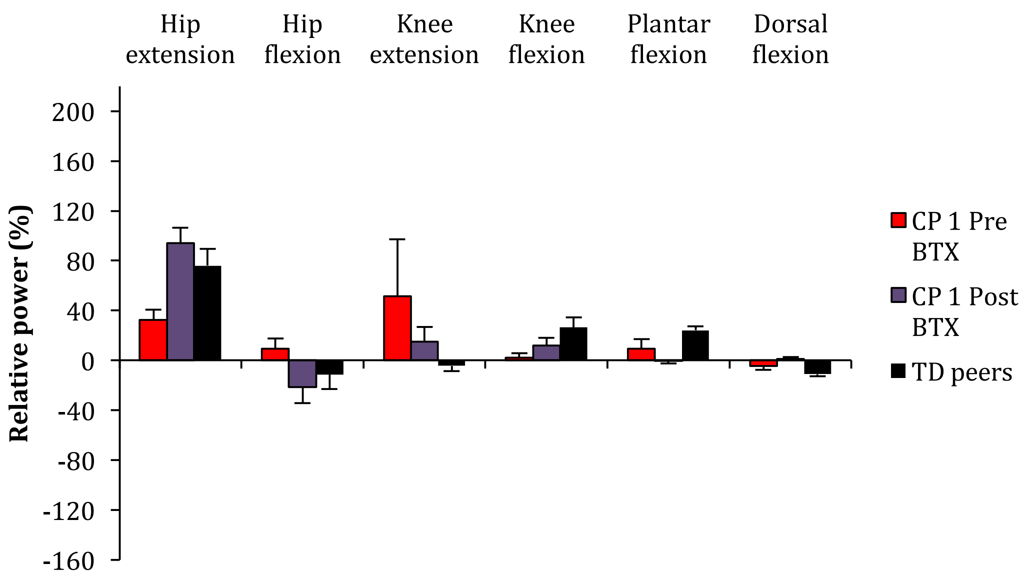


Figure A183: Joint powers produced at 50 RPM by treated (top) and non-treated (bottom) leg.

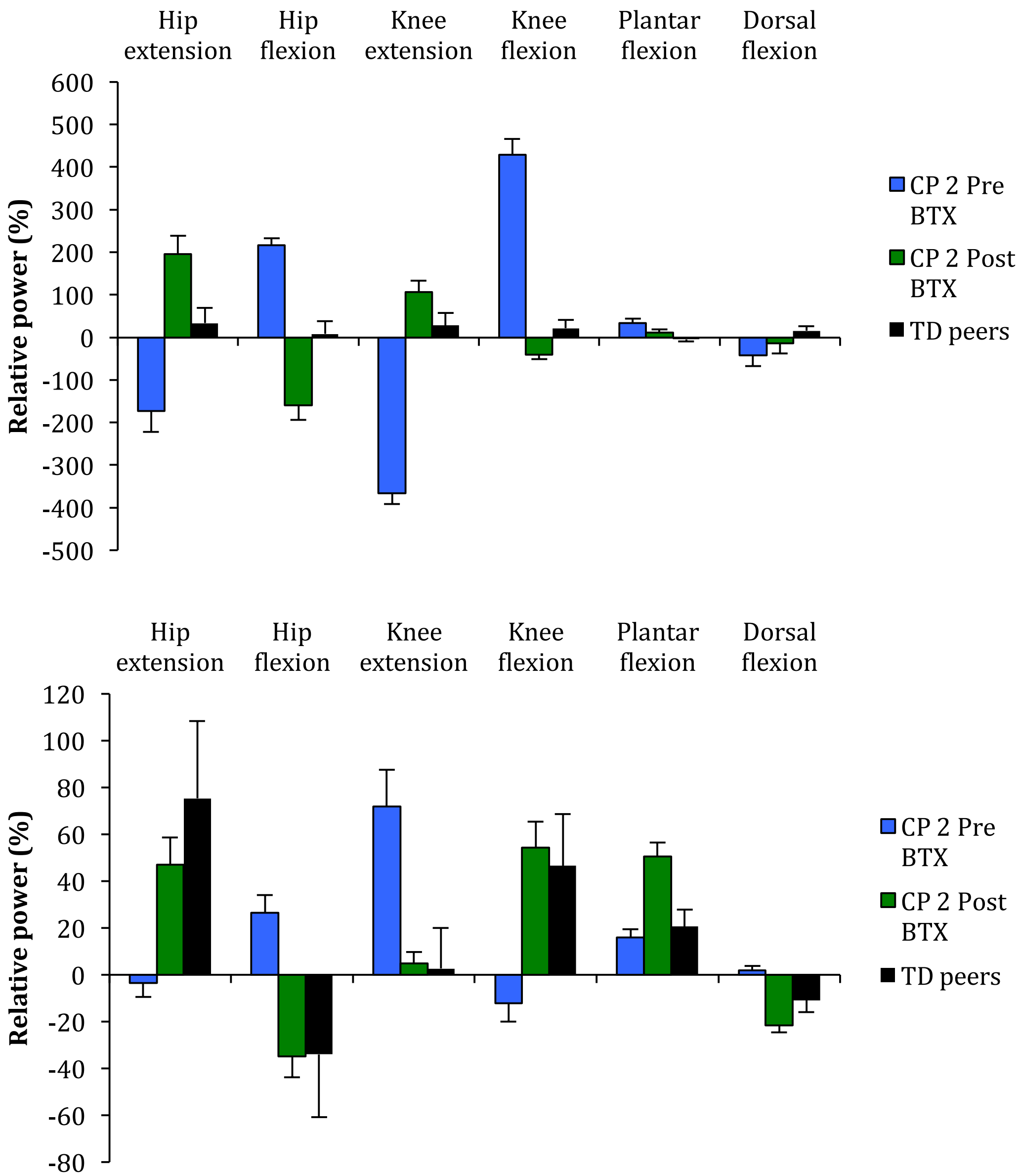


Figure A184: Joint powers produced at 60 RPM by treated (top) and non-treated (bottom) leg.

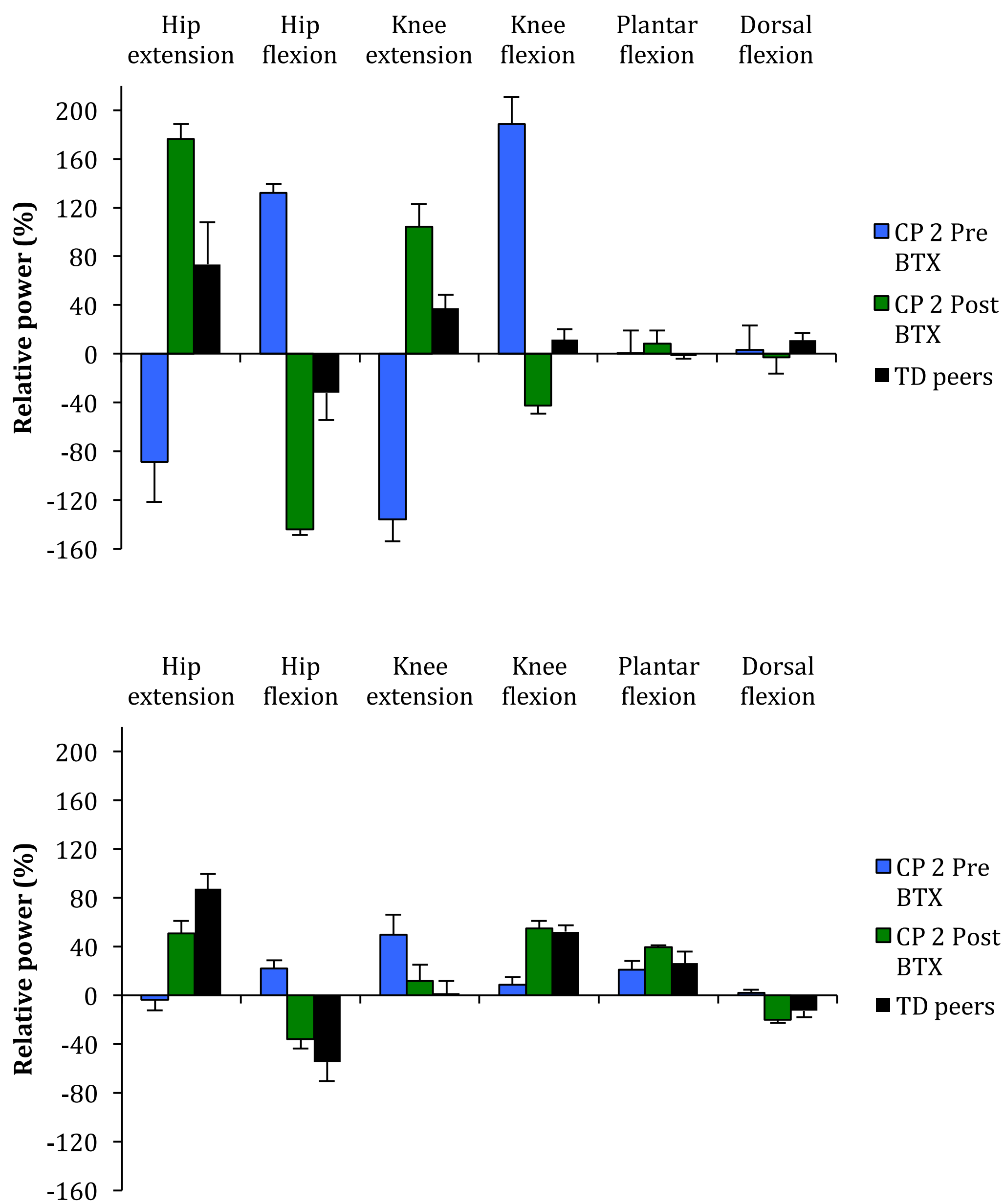


Figure A185: Joint powers produced at 70 RPM by treated (top) and non-treated (bottom) leg.

Abbreviations of terms

ASIS = anterior superior iliac spine

BDC = bottom dead center

BTX = botulinum toxin

CP = cerebral palsy

GAS = gastrocnemius

GMFCS = Gross Motor Function Classification Scale

HS = hamstring

MAS = Modified Ashworth Scale

non-Tx = non-treated

ROM = range of motion

RPM = revolutions per minute

TD = typically developing

TDC = top dead center

Tx = treated

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