LONG-TERM FOLLOW-UP OF CHILDREN WITH IDIOPATHIC TOE WALKING

by

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ABSTRACT

Introduction: Toe walking may develop with early ambulation, but is considered abnormal in healthy children after three years of age. Idiopathic toe walking is a diagnosis of exclusion characterized by forefoot weight bearing and lack of heel contact with the floor. This condition may be associated with pain in the legs or feet, frequent tripping or falling, and ankle injuries. In British Columbia, treatment for toe walking varies from physiotherapy, casts or braces, Botulinum toxin A injections into calf muscles, and/or surgery. Little evidence exists regarding long-term treatment effects.

Objectives: To determine the natural history of idiopathic toe walking and the long-term impact of treatment on gait outcomes and severity of toe walking. Potential activity limitations and participation restrictions were also explored.

Methods: Gait analysis data and severity of toe walking were compared from baseline to follow-up in 44 adolescents and young adults diagnosed with idiopathic toe walking between 1997 and 2005 in a non-experimental retrospective cohort study. Participants were grouped as receiving no treatment other than stretching exercises (n=20) or treatment (n=24), including casting +/- Botulinum toxin A injections (n=23) or tendo-Achilles lengthening (n=1). Levels of activity and participation were assessed using the Pediatric Outcomes Data Collection Instrument for adolescents and the Medical Outcomes Study 36-Item Short Form Survey Instrument for young adults.

Results: Ankle kinematics improved in the treatment group only whereas improved ankle kinetics and compensatory knee hyperextension were observed in both groups. Ankle moments remained atypical in 89% of the participants at follow-up. Clinical ankle dorsiflexion decreased

over time in both groups. Statistically significant differences in severity at follow-up were found only in the treatment group ($p<0.001$). Adolescents and young adults scored 54% and 68%, respectively, above the general population mean in global functioning and physical capacity by self-report.

Conclusions: The natural history of idiopathic toe walking indicates improved timing of ankle kinematics and improved ankle kinetics. Participants treated for toe walking as children demonstrated significant changes in kinematics, kinetics, and severity at follow-up. Despite perseverance in gait changes in both groups at follow-up, self-report questionnaires suggest there is minimal impact on activity and participation.

PREFACE

The master's candidate designed and implemented this study with guidance from the supervisory committee: Dr. Liisa Holsti, Dr. Michael Hunt, and Mr. Alec Black. The study and corresponding methods were approved by the University of British Columbia Children's and Women's Research Ethics Board (certificate number H15-00556). Dr. Robbin Hickman served as an additional resource and provided input into the methodology required for the American Academy for Cerebral Palsy and Developmental Medicine systematic review included in Chapter Two. Ms. Karen Sauve was involved as the second reviewer required for completion of the systematic review. The master's candidate collected all data with assistance from Mr. Black and performed analyses with guidance from Mr. Boris Kuzeljevic and Dr. Bruno Zumbo. This thesis manuscript was written in full by the master's candidate with editing contributions from Dr. Holsti.

A version of the Prevalence of Idiopathic Toe Walking, Normal Function of the Foot and Ankle, and Proposed Theories of Etiology of Idiopathic Toe Walking information in Chapter Two and the Gait Analysis Data information in Chapter Three has been submitted as an invited chapter. Davies, K., Leveille, L., and Alvarez, C. (2016). *Idiopathic Toe Walking.* In: Müller, B. and Wolf, S. (Eds.). Handbook of Human Motion.

Use of the Pediatric Outcomes Data Collection Instrument for the purpose of this study was available without copyright restrictions or registration. The license agreement for use of the Medical Outcomes Study 36-Item Short Form Survey Instrument is included in the relevant appendix.

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LIST OF ABBREVIATIONS

AACPDM……American Academy for Cerebral Palsy and Developmental Medicine

AFO……...……Ankle Foot Orthotic

AM1……...……...……First Ankle Moment

AM2……...……...Second Ankle Moment

ANOVA……...…Analysis of Variance

BC……..British Columbia

BMI……Body Mass Index

BoNT-A……...Botulinum toxin A

CI……Confidence Interval

CP……...……Cerebral Palsy

CPA…….Canadian Physiotherapy Association

DF……...……...Dorsiflexion

EMG……..…..Electromyography

FO……Foot Orthotic

ICC……...Intraclass Correlation Coefficient

ICF-CY....International Classification of Functioning, Disability & Health for Children & Youth

ITW…….Idiopathic Toe Walking

NT…….Non-Treatment or No Treatment

OT……..Occupational Therapy

PROM……...Passive Range of Motion

PDMS-2……..Peabody Developmental Motor Scales - Revised

PF……...Plantarflexion

PODCI…… Pediatric Outcomes Data Collection Instrument

PROM……...Passive Range of Motion

PT……..….Physical Therapy

SD……...Standard Deviation

SF-36……Medical Outcomes Study 36-Item Short Form Survey Instrument

SEM……...Standard Error of Measurement

SGL……..Shriners Gait Lab

SSRD…….Single Subject Research Design

SHHCC……Sunny Hill Health Centre for Children

RCT……Randomized Control Trial

ROM……..Range of Motion

TAL……..…Tendo-Achilles Lengthening

WHO……..World Health Organization

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I owe specific thanks to Barbara Kelly for taking the time to peer review our systematic review protocol and provide thoughtful feedback; to Karen Sauve who dedicated hours and late nights to systematically reviewing literature related to this thesis; to Jessica Maurer who patiently assisted and talked me through the data extraction process and any questions I had pertaining to biomechanics; to Tara Klassen whose questions taught me to think more deeply about my own questions, and to Val Ward, who supported me unequivocally.

I truly value the support I have received from the rest of my friends, family, fellow graduate students, and colleagues at Sunny Hill Health Centre for Children – your genuine interest in my progress and this thesis project kept me going!

It was a privilege to work with all the participating adolescents, young adults, and their families throughout the recruitment and data collection phases of this project. I would like to express my sincere gratitude for the time and effort you contributed to this research.

Lastly, I am grateful for the funding I received for this research project from the University of British Columbia and the Canadian Institutes of Health Research.

To my children

1. INTRODUCTION

Idiopathic toe walking (ITW) is a condition characterized by a persistent bilateral toe-toe gait pattern, absent or diminished heel contact with the floor, and no other identified underlying diagnoses. Toe walking may develop with independent walking or within the first year of independent ambulation, but is considered abnormal in typically developing children after the age of two to three years (Sutherland, Olsen, Cooper & Woo, 1980; Engstrom & Tedroff, 2012; Sobel, Caselli & Velez, 1997). Heel strike at initial contact is typically present by 18 months of age or within a range of three to 50 weeks following the onset of independent walking (Sutherland et al., 1980, Burnett & Johnson, 1971). The differential diagnoses associated with ITW include cerebral palsy, spinal cord abnormalities, myopathy, peripheral neuropathy, neuromuscular disorders, dystonia, ankylosing spondylitis, autistic spectrum disorders, schizophrenia, talipes equinovarus, leg length discrepancy, venous malformation or tumour in the gastrocnemius muscle, or trauma (Hicks, Durinick & Gage, 1988; Shulman, Sala, Chu, McCaul & Sandler, 1997; Le Cras, Bouck, Brausch & Taylor-Haas, 2011). In 1967, Hall, Salter, and Bhalla (1967) were the first to describe persistent toe walking in children and diagnosed this condition as congenital short tendo calcaneus in the absence of other known pathology. Subsequently, this condition has been identified as toe walking, habitual toe walking and hereditary tendo Achilles contractures, reflecting a difference in opinion regarding the etiology of ITW (Sobel et al., 1997; Hall et al., 1967; Crenna, Redrizzi, Andreucci, Frigo & Bono, 2005; Katz & Mubarak, 1984).

The lack of a clear etiology for ITW makes it difficult to determine whether treatment needs to target toe walking, more global sensory issues, or if it only has an impact on ankle range of motion. Few studies explore the natural history of ITW and the history of toe walking

following treatment which limits the evidence for appropriate management of this condition. Thus, the purpose of this study is to explore the natural history of ITW and the impact of treatment of ITW on gait outcomes.

Chapter Two of this thesis provides a literature review to outline the known prevalence, etiology, and proposed theoretical constructs of ITW, as well as a systematic review of the evidence of current treatment approaches for children with ITW. The systematic review considers the effects of intervention on outcomes representing all components of functioning for a child. This review of the current literature provides the rationale for the proposed research study.

Chapter Three describes the methods used for this study including recruitment strategies and the inclusion and exclusion criteria of the population sample. Described further are the protocol and details of the outcome measures as well as the data synthesis and analysis used to explore the three objectives of this study.

Chapter Four provides the results related to the objectives of the study. Statistical significance is reported where relevant and results from data used to calculate group differences are reviewed separately from descriptive data. Tables and figures provide a coherent summary of the primary outcome measures.

Lastly, Chapter Five presents a discussion which describes and clarifies the clinical significance and interpretation of the study results. The findings are considered in light of present and pertinent literature surrounding ITW in relation to the study hypotheses. Study limitations and strengths are reviewed as well as recommendations for future research and clinical practice.

2. LITERATURE REVIEW[1]

2.1 Prevalence of Idiopathic Toe Walking

Currently, the best estimates are that ITW occurs in 4.9 to 24% of all children (Engström & Tedroff, 2012; Sobel et al., 1997; Accardo, Morrow, Heaney, Whitman & Tomazic, 1992). The most recent study examined a large cohort of 1,436 children and found that the prevalence rate decreased to 2.1% by 5.5 years of age (Engström & Tedroff, 2012). There is a higher frequency of ITW reported in males compared to females (Engström & Tedroff, 2012; Fox, Deakin, Pettigrew & Paton, 2006; Eastwood, Memelaus, Dickens, Broughton & Cole, 2000; Stricker & Angulo, 2000). In addition, a positive family history has an overall reported incidence of 10-40% for both males and females (Engström & Tedroff, 2012; Sobel et al., 1997; Fox et al., 2006; Stricker & Angulo, 1998). A recent observational study of 836 children with ITW found 64% were males (χ^2<0.001) and 42% presented with a positive family history (χ^2<0.001), also suggestive of a genetic component with males affected more than females (Pomarino, Ramirez Llamas, & Pomarino, 2016). The broad range in reported prevalence is a result of differing defining characteristics of ITW in these studies and the length of follow-up data. In part, it is difficult to determine the true prevalence of ITW given the number of gait deviations which develop as a result of forefoot weight bearing and a lack of normal ankle range of motion.

2.2 Normal Function of the Foot and Ankle

As part of normal gait, the foot acts as a stable platform, there is appropriate muscular power generation and absorption, the knee and ankle joints are in a plane of progression, and

[1] A version of the Prevalence of Idiopathic Toe Walking, Normal Function of the Foot and Ankle, and Proposed Theories of Etiology of Idiopathic Toe Walking information in Chapter Two has been submitted as an invited chapter. Davies, K., Leveille, L., and Alvarez, C., (2016). *Idiopathic Toe Walking*. In: Müller, B. & Wolf, S. (Eds.). Handbook of Human Motion.

there is adequate range of motion (ROM) at the ankle. During a mature heel-toe gait, ankle motion averages through an arc of neutral to 10° of dorsiflexion (DF) and neutral to 20° of plantarflexion (PF) (Perry, 1992). Typically developing children are born with approximately 54° of ankle DF passive range of motion (PROM), decreasing to 41° PROM at two years (Walker, 1991), 20° to 30° PROM by four to seven years of age (Cusick & Stuberg, 1992), an average of 27° PROM in nine to 13 year olds and 27.5° PROM in 14-17 year olds (Grimston, Nigg, Hanley & Engsberg, 1993). The normative range of passive ankle DF reported for young adults is 8° to 25° (Grimston et al, 1993; Bovens, van Baak, Vrencken, Wijnen & Verstappin, 1990). Ankle position contributes to each of the determinants of normal gait, as described by Perry (1985) and Gage (2004), including stability in stance, foot clearance, pre-positioning the foot in swing, appropriate step length and energy conservation. It is suggested that children learn to walk instinctively without these determinants and are reliant on gradual central nervous system maturation to attain typical heel-toe progression (Sutherland et al., 1980; Sutherland, Olsen, Biden, & Wyatt, 1988; Gage, 2004).

Perry (1992) divides normal gait into periods of initial contact, loading response, mid-stance, terminal stance, pre-swing, mid-swing, and terminal swing. Further, the ankle and foot action during stance is also described in terms of three rockers: the first (heel) rocker occurs from foot contact to foot flat with controlled PF, the second (ankle) rocker occurs when the tibia moves over the foot in stance, and the third (forefoot) rocker occurs when the foot moves into PF again for push-off. The first rocker begins with foot contact and continues to approximately 8% of the gait cycle, overlapping slightly with the second rocker, which takes place from about 5% to 45% of the gait cycle. The second rocker also overlaps with the third rocker, described by Perry (1992) as starting with heel lift at approximately 30% of the gait cycle and continuing until

the termination of stance. Baker (2014) proposes that the third rocker may even start as early as 20% of the gait cycle.

Children with ITW often demonstrate equinus pre-positioning in late swing leading to an absent first rocker and/or shortened loading response with progressive ankle DF following initial foot contact instead of the controlled PF typically observed (Crenna et al., 2005; Hicks et al., 1988; Kelly, Jenkinson, Stephens, & O-Brien, 1997). Diminished ankle DF ROM during gait limits the forward progression of the tibia over the foot and a reversal of the second rocker, resulting in an abnormal and early transition from DF to PF (Hicks et al., 1988). This is also described as an early third rocker by Alvarez, De Vera, Beauchamp, Ward, and Black (2007), referring to premature progression into PF at or before 30% of stance phase. Clinically, the lack of normal ankle motion during gait presents as a toe strike or a flat foot at initial contact and early heel rise may be present prior to mid-stance. Other reported gait deviations in ITW include an increased anterior pelvic tilt, mild knee hyperextension, and external foot progression (out-toeing) as well as abnormal loading of the foot and absent body weight transfer during mid-stance (Hicks et al., 1988; Stott, Walt, Lobb, Reynolds, & Nicol, 2004; Westberry, Davids, Davis, & de Morais Filho, 2008; McMulkin, Baird, Caskey, & Ferguson, 2006; Clark, Sweeney, Yocum, & McCoy, 2010). Unlike other gait pathology, children with ITW demonstrate variable patterns with the ability to spontaneously alternate between bilateral toe-toe and heel-toe gait, irrespective of normal or limited ankle DF ROM (Hicks et al., 1988), and the origin of ITW remains unclear.

2.3 Proposed Theories of Etiology of Idiopathic Toe Walking

There are a number of proposed biomechanical, motor control, and neurological theories behind the etiology of ITW. It has been suggested that toe walking is a result of congenital contractures of the gastrocnemius-soleus muscle complex; however, the presence of equinus contractures at birth with ITW has not been confirmed in the literature. Older children and exclusive toe walkers typically demonstrate more restrictions in ankle dorsiflexion than younger children or intermittent toe walkers (Sobel et al., 1997; Hall et al., 1967; Stricker & Angulo, 1998); alternatively suggesting that limited DF and ITW is a result of increased time spent walking on toes (Furrer & Deonna, 1982). Histological analysis demonstrates a high proportion of Type one muscle fibres in gastrocnemius in children with ITW, although this may also be a result of adaptive changes from prolonged toe walking (Eastwood, Dennett, Shield, & Dickens, 1997). Biomechanical theory is useful but not definitive and Clark et al. (2010) proposed that immature motor control may be present in children with persistent ITW. Clark et al. (2010) postulated that delayed achievement of both an erect posture and reciprocal activation between dorsiflexors and plantarflexors causes forward trunk alignment with resultant forefoot weight bearing and absent heel strike, suggestive of incomplete sensory-motor development.

It is possible, given family history studies and absence of typical response to treatment, that genetic factors, subtle neurological changes or impairment could underlie ITW (Eastwood et al., 1997; Stricker & Angulo, 1998). Studies have reported a positive association between language delays, learning disabilities, prematurity, and ITW (Accardo et al., 1992; Fox et al., 2006; Stricker & Angulo, 1998). The incidence of toe walking reported in children with autism or pervasive developmental disorders is 19-21% (Ming, Brimacombe, & Wagner, 2007; Barrow, Jaworski, & Accardo, 2011). A study of children aged 19-36 months with recently diagnosed

autism demonstrated no association between toe walking and sensory dysfunction or language delays; yet a significant association with persistent components of the tonic labyrinthine reflex was found, suggestive of the contribution of a motor deficit (Accardo & Barrow, 2015). Engström, Bartonek, Tedroff, Orefelt, Haglund-Åkerlind, and Gutierrez-Farewik (2013) investigated whether an increased prevalence of neuropsychiatric symptoms had a negative influence on treatment results – and they did not, although their study was inconclusive as a result of a small population sample.

Dynamic electromyography (EMG) data of children with ITW have shown atypical co-contraction and out of phase muscle activity with early firing of gastrocnemius in swing and low amplitude firing of tibialis anterior through parts of stance and swing (Griffin, Wheelhouse, Shiavi, & Bass, 1977). Another gait EMG study demonstrated similar findings in children with toe walking, cerebral palsy, and equinus deformities compared to a matched control group of children with typical gait asked to walk on their toes (Kalen, Adler, & Bleck, 1986). Brunt, Woo, Kim, Ko, Senesac, and Li (2004) showed that early gastrocnemius activity in swing phase and early and limited duration tibialis anterior activity is likely to result in flatfoot or forefoot weight bearing. Despite the fact that these findings are suggestive of a possible central nervous system mechanism, a biomechanical etiology of tendo-Achilles contracture cannot be ruled out. EMG studies show high variability and are not necessarily conclusive in providing objective diagnoses due to inconsistent findings (Griffin et al., 1977; Papariello & Skinner, 1985; Kalen et al., 1986).

A study investigating sensory processing and motor skill abilities in four-to-eight year old healthy children with and without ITW showed that children with ITW have both sensory and motor challenges (Williams, Tinley, Curtin, Wakefield, & Nielsen, 2014). These children processed sensory input differently than the children without ITW and Williams, Tinley, Curtin,

and Nielsen (2012) also found that children with ITW demonstrated diminished vibration perception thresholds. The motor and sensory challenges may be linked with ITW, but they are not strong enough to indicate distinct clinical diagnoses and the etiology of ITW is still unclear.

The reported clinical manifestations of ITW include pain in the legs or feet, frequent tripping or falling, poor balance, foot-wear problems, and limitation in PROM of ankle DF (Sobel et al., 1997; Engelbert, Gorter, Uiterwaal, van de Putte, & Helders, 2011; Fox et al., 2006; Clark et al., 2010; Hirsch & Wagner, 2004). The latter is associated with ankle injuries as well as forefoot, midfoot, and/or hind foot pathology (Tabrizi, McIntyre, Quesnel, & Howard, 2000; DiGiovanni et al., 2002; Hill, 1995). Clinical measurement of ankle DF PROM is not correlated with maximum ankle DF in stance, as determined by three-dimensional gait analysis studies (Alvarez et al., 2007; Stott et al., 2004; McMulkin, Gordon, Tompkins, Caskey, & Baird, 2016), making recommendations for treatment challenging. From the perspective of the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY), these manifestations of ITW are conceptualized as impairments in body structures and body functions and may cause restrictions in daily function (World Health Organization, 2007; International Classification of Functioning, Disability and Health, 2013), indicating a need for intervention.

2.4 Current Treatment Approaches for Idiopathic Toe Walking

In British Columbia (BC), management options for ITW typically include physical therapy, orthotics, casting (with or without BoNT-A injections to the gastrocnemius and soleus muscles), and/or surgery. ITW clinical care guidelines recommend an intervention and treatment frequency algorithm including several physical therapy (PT) modalities: stretching, strengthening, manual therapy, balance and coordination exercises, gait training, and home

exercise programs (Le Cras et al., 2011). Further, a systematic review comparing casting and surgical treatment in children with ITW found surgery to be superior for improving ankle DF PROM; however, neither surgical nor non-surgical treatment differed significantly in terms of persistent toe walking (van Bemmel, van de Graaf, van den Bekerom, & Vergroesen, 2014). Another systematic review, published in the same year, extended this opinion by adding that only the effects of surgery are reported as lasting more than one year, without consideration for the effect of treatment on activities and participation (van Kuijk, Kosters, Vugts, & Geurts, 2014). Although van Bemmel et al. (2014) included studies with greater rigour (i.e., evidence levels I through IV), their review lacked a description of the Level of Evidence Scoring system, making it difficult to fully appreciate their results.

There are significant costs associated with both the conservative and non-conservative treatment of ITW, such as financial costs and potential pain and time in rehabilitation for the child. Moreover, irrespective of approach, treatment has significant health system costs and greater clarity is needed regarding best practice for assessment and management of ITW. At the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) 2014 Annual Meeting, another treatment algorithm for ITW was presented involving a conservative approach focused on serial casting, orthotics, and PT with a component of sensory assessment with referral to Occupational Therapy (OT) as indicated (Maus et al., 2014). A protocol by Williams, Michalitsis, Murphy, Rawicki, and Haines (2013) proposed to examine the impact of foot wear, carbon fibre orthotics, and whole-body vibration on gait parameters in this population with recent study results suggesting a short-term improvement in gait as a result of whole-body vibration (Williams, Michalitsis, Murphy, Rawicki, & Haines, 2016). The focus continues to

target the domains of body structures and functions, highlighting the ongoing need to examine the impact of these interventions, if any, on the domain of activity and participation.

To update and to evaluate the outcome of treatment strategies on the management of children with ITW using the ICF framework to capture the impact of intervention on all domains, the AACPDM methodology was used to evaluate and to describe the literature systematically (Darrah, Hickman, O'Donnell, Vogtle, & Wiart, 2008; Wiart et al., 2012). Further coding of treatment outcomes into ICF-CY components was done to summarize the present state of the evidence, providing rationale to support the aims of the proposed research project. These study objectives will be described below.

The AACPDM systematic review methodology was developed to summarize the research literature related to specific treatment strategies for children with developmental disabilities (Darrah et al., 2008). This methodology codes levels of evidence and assesses study quality for group intervention, single subject design, and systematic review studies. Furthermore, it evaluates treatment outcomes from an ICF framework, providing possible links between effects and components of the ICF.

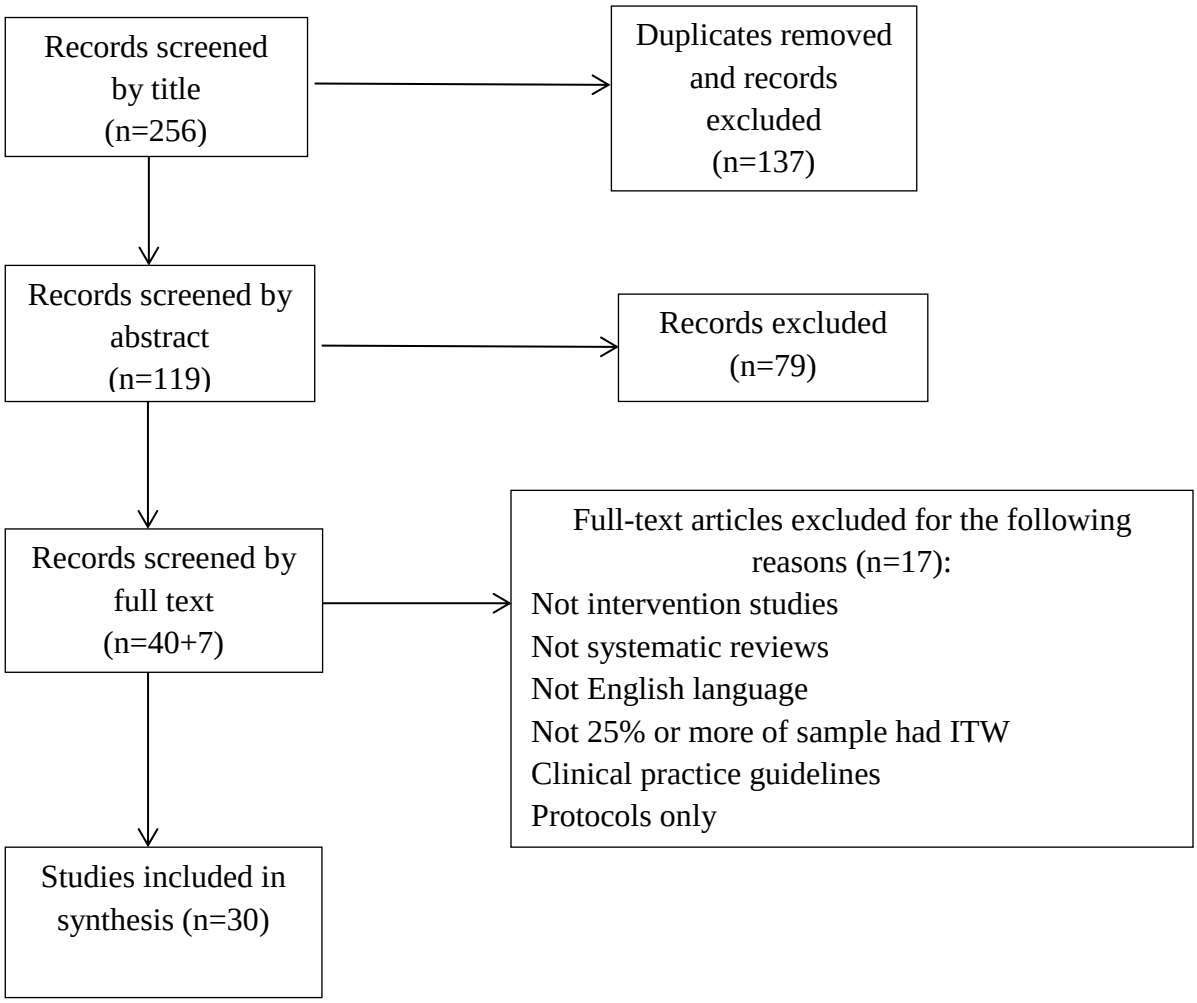
2.4.1 Search Strategy

Methods for study selection, data collection, and analysis were identified and peer-reviewed in advance based on recommendations from the PRISMA statement and the Cochrane Collaboration (Liberati et al., 2009; Higgins & Green, 2011). The following electronic databases were searched for relevant studies: Cumulative Index to Nursing and Allied Health Literature (CINAHL) 1982 to June 2014; Cochrane Database of Systematic Reviews 1993 to June 2014; Cochrane Central Register of Controlled Trials; Database of Reviews of Effectiveness (DARE); EMBASE 1946 to June 2014 (Ovid); ERIC 1969 to June 2014 (EBSCO); Google Scholar;

MEDLINE 1946 to June 2014 (Ovid); OTSeeker; Physiotherapy Evidence Database (PEDro); PsycINFO (EBSCO); PubMED; and Web of Science. Electronic search terms for study identification included a combination of 'gait', 'toes', 'gait disorders', 'equinus contracture or deformity', 'idiopathic toe walking', 'habitual toe walking', 'serial or surgical casting', 'foot orthoses', 'physiotherapy', 'botulinum toxins type A', 'Achilles tendon' or 'gastrocnemius or triceps surae or heel cord lengthening', with no initial limits set for study design or language of publication. Additional studies were retrieved by manually searching reference lists of included studies and by hand searching Developmental Medicine and Child Neurology, the Journal of Pediatric Orthopaedics, and Gait and Posture from 2012 to 2014. The literature search was updated in June 2016. The review examined only original, peer-reviewed literature published in scientific journals. The complete search strategy is available by request.

There were 256 articles identified from the initial database search. After duplicates were removed and records were screened by title and abstract, 40 articles were reviewed by full text and 17 studies were excluded based on the inclusion and exclusion criteria. Following the updated literature search in June 2016, seven additional studies were found, leaving 30 articles for analysis (Figure 2).

Figure 2.1. Literature Search Flow Diagram



2.4.2 Inclusion and Exclusion Criteria

Articles were included for review and analysis if: (1) they were randomized control trials (RCTs), systematic reviews, and group and single participant research design studies, (2) participants were children up to age 18 with a diagnosis of ITW, and (3) they involved surgical intervention or conservative treatment including physical therapy, serial casting, BoNT-A injections, orthotic interventions, or any other intervention with a valid outcome measure provided by an allied health or medical professional. Articles were excluded if they were: (1) not

intervention studies, (2) not systematic reviews, (3) not written in the English language, (4) clinical practice guidelines, (5) protocols only, or (6) a sample of children with ITW constituting less than 25% of the study population.

2.4.3 Data Extraction and Organization

Included studies were screened by two independent reviewers to determine if inclusion criteria were met. They then assessed methodological rigour and extracted data using data extraction sheets based on the AACPDM's recommendations (Darrah et al., 2008). Data extraction sheets were pilot tested on two randomly-selected included articles. Extracted data were summarized in a table and: (1) assigned a level of evidence based on classification by the Oxford Centre for Evidence Based Medicine (OCEBM Levels of Evidence Working Group, 2011) (Tables 2.1a, 2.1b, 2.2); (2) appraised for study quality to address risk of bias (Tables 2.3a, 2.3b, 2.3c), and; (3) coded by the representative ICF component for treatment outcomes (Table 2.4). There was unanimous consensus on study inclusion and any disagreements regarding levels of evidence or study conduct were resolved by discussion and referred to a third party as necessary. It was not necessary to contact authors to gain further information.

Table 2.1a. Levels of Evidence for Group Designs

Level	Intervention (Group) Studies
I	Systematic review of randomized controlled trials (RCTs) Large RCT with narrow confidence intervals) (n>100)
II	Smaller RCT's (with wider confidence intervals) (n<100) Systematic reviews of cohort studies "Outcomes research" (very large ecologic studies)
III	Cohort studies (must have concurrent control group) Systematic reviews of case control studies
IV	Case series Cohort study without concurrent control group (e.g. with historical control group) Case-control study
V	Expert opinion Case study or report Bench research Expert opinion based on theory or physiologic research Common sense/anecdotes

Table 2.1b. Levels of Evidence for Single Subject Research Designs

Level	Single Subject Research Design Studies (SSRD)
I	Randomized controlled *n*-of-1 (RCT), alternating treatment design (ATD), and concurrent or non-concurrent multiple baseline design (MBDs); generalizability if the ATD is replicated across three or more subjects and the MBD consists of a minimum of three subjects, behaviours, or settings. These designs can provide causal inferences.
II	Non-randomized, controlled, concurrent MBD; generalizability if design consists of a minimum of three subjects, behaviors, or settings. Limited causal inferences.
III	Non-randomized, non-concurrent, controlled MBD; generalizability if design consists of a minimum of three subjects, behaviors, or settings. Limited causal inferences.
IV	Non-randomized, controlled SSRDs with at least three phases (ABA, ABAB, BAB, etc.); generalizability if replicated across three or more different subjects. Only hints at causal inferences.
V	Non-randomized controlled AB SSRD; generalizability if replicated across three or more different subjects. Suggests causal inferences allowing for testing of ideas.

2.4.4 Study Types, Participants and Interventions

This review includes three group research designs of level II evidence, with one conduct rating of moderate and two of weak quality, and two level III studies, both of weak quality (Table 2.2). One single subject design study was included and rated as level three evidence of moderate quality. Two systematic reviews were included and both rated as level II evidence of moderate quality (van Bemmel et al., 2014; van Kuijk et al., 2014). Additionally, 19 studies contributed level IV evidence and three studies added level V evidence. One such study was the original work introducing the condition of ITW in 1967 (Hall et al., 1967). These level IV and V studies were not sufficiently robust to be included in the final evidence table for study conduct (Table 2.4). However, these studies were included in the discussion of adverse events and study strength so as to highlight the present state of the evidence for ITW treatment.

The intervention studies reviewed included a total of 616 children with ITW. The sample also included 41 children with cerebral palsy (CP), five children with other diagnoses, and 15 children described as typically developing. The age of the children ranged from 18 months to 17 years at the time of initial evaluation. Sex was reported in 26 intervention studies including 392 boys and 262 girls. Most of these studies lacked sufficient reporting on functional ability levels for their samples. Heterogeneity was found across study type, participant sample, and reporting of ITW severity. In an attempt to account for this heterogeneity, the systematic review by van Bemmel et al. (2014) used differences in mean patient ages for their sample of 298 children. The primary emphasis of ITW interventions was to improve ankle DF PROM and decrease time spent toe walking. Few studies considered activity levels, participation, or contextual components of the ICF (Stott et al., 2004; Clark et al., 2010; Kogan & Smith, 2001; Jacks,

Michels, Smith, Koman, & Shilt, 2004; van Bemmel, van den Bekerom, Verhart, & Vergroesen, 2012; Pistilli, Rice, Pergami, & Mandich, 2014; McMulkin et al., 2016; Sätilä et al., 2016).

Table 2.2. Summary of Studies: Interventions and Participants

Group Studies: Citations	Level of Evidence, Conduct Rating and Research Design	Participants	Total *n*	Ages	Intervention
Hall et al. (1967)	V Case report; Prospective	Children with contractures of calf muscles and toe walking	*n*=20 3 CP 13 ITW 4 other	Mean 7.5y	Sx: calcaneal tendon lengthening + BK cast x 6wks (3wks NWB, 3 wks WB) Ctl: None
Griffin et al. (1977)	IV Case series; Prospective	Children with limited DF and habitual toe walking	*n*=6	5-9y	Cast: 6wks in serial casts with max DF + DF ex's + heel-toe gait training post-casting Ctl: None
Conrad & Bleck (1980)	IV Case series; Prospective	Children with CP and ITW with dynamic equinus	*n*=8 6 CP 2 ITW	3-9y	Augmented auditory feed-back: heel sensor x 1hr/day x 1-6 mo (mean 3mo) Ctl: None
Katz & Mubarak (1984)	IV Case series; Prospective	Children with Achilles tendon contractures, toe walking	*n*=8	3-10y	Serial cast: DF cut-out casts permitting active DF, ex's + negative heel shoes x 2-16wks Ctl: None
Stricker & Angulo (1998)	III-W (2/7) Cohort study with concurrent control group; Retrospective	Children with ITW	*n*=80 48 Ctl 17 CA 15 Sx	2-13y at initial ax	Cast/Orthotics: 6-12wks in BK casts or 3-8mo in solid AFOs + stretching post-cast/AFO Sx: bilat open Achilles z-plasty lengthening or bilateral GM recession + AFOs 2-6mo post-op Ctl: observation, special shoes or heel cord stretch ex's with PT/parents

Group Studies: Citations	Level of Evidence, Conduct Rating and Research Design	Participants	Total *n*	Ages	Intervention
Eastwood et al. (2000)	III-W (3/7) Cohort study with concurrent control group; Retrospective	Children with ITW	*n*=136 49 Ctl 41 CA 46 TAL	18mo -13y	Serial cast: 6wks in bilat BK walking casts Sx: Baker type aponeurotic TAL + BK walking casts x 6wks post-op Ctl: observation
Brouwer, Davidson, & Olney (2000)	IV Cohort study without concurrent control group; Prospective	Children with CP and ITW	*n*=16 8 CP 8 ITW	3-12y	Cast: 3-6wks in BK walking casts, changes at 1-2wks Ctl: None
Kogan & Smith (2001)	IV Cohort study without concurrent control group; Retrospective	Children with ITW	*n*=10	Not reported	Sx: percutaneous TAL + BK walking cast x 4wks post-op. Ctl: None
Stott et al. (2004)	IV Cohort study without concurrent control group; Retrospective	Adolescents and young adults treated for ITW	*n*=13	16-25y	Cast: 6wks, 3 sets of changes + stretching ex's post-casting Sx: serial casting x 6wks + stretching + percutaneous TAL or Baker's GM-SOL lengthening Ctl: None
Hirsch & Wagner (2004)	IV Cohort study without concurrent control group; Retrospective	Children with ITW 7-21y post-initial PT ax	*n*=14	13-28y	PT: passive GM-SOL stretching +/- DF strengthening ex's Cast: BK 2-4wk + PT +/- dynamic night splints post-cast removal Ctl: None
Jacks et al. (2004)	IV Case series; Retrospective	Children with ITW	*n*=10	2-17y	Cast + BoNT-A: injection to GM/SOL followed by 1-3wks BK walking casts, then AAFOs + home stretching + strengthening program Ctl: None

Group Studies: Citations	Level of Evidence, Conduct Rating and Research Design	Participants	Total *n*	Ages	Intervention
Brunt et al. (2004)	IV Cohort study without concurrent control group; Prospective	Children with persistent toe walking	*n*=5	3-6y	BoNT-A: injection to GM/SOL, followed by PT 2x/wk for gait training + ex's to ↑ functional use of TA + DF ROM + home program Ctl: None
Hemo, Macdessi, Pierce, Aiona, & Sussman (2006)	IV Cohort study without concurrent control group; Retrospective	Children with ITW who failed non-operative treatments	*n*=15	4-13y	Sx: bilat open/percutaneous TAL, then BK walking cast 4-6wks, then AFO fulltime progressing to nighttime, then d/c Ctl: None
McMulkin et al. (2006)	IV Cohort study without concurrent control group; Retrospective	Children with ITW + history of surgical lengthening	*n*=14	5-12y	Sx: previous Vulpius-type GM lengthening or percutaneous TAL Ctl: None
Fox et al. (2006)	IV Cohort study without concurrent control group; Prospective	Children with ITW	*n*=44	2-14y	Cast: BK walking casts in plantigrade, changes at 2wk intervals for 3-10wks, home program for passive Achilles tendon stretching. Ctl: None
Jahn, Masavada, & McMulkin (2009)	IV Cohort study without concurrent control group; Retrospective	Children with equinus gait in CP + ITW	*n*=38 24 CP 14 ITW	3-15y	Sx: previous TAL or Vulpius procedure Ctl: None

Group Studies: Citations	Level of Evidence, Conduct Rating and Research Design	Participants	Total *n*	Ages	Intervention
Grady & Kelly (2010)	IV Cohort study without concurrent control group; Prospective	Children with equinus from ITW	*n*=22	7-16y	Sx: endoscopic GM recession following at least 6mo of unsuccessful conservative treatment (NSAIDS, PT, stretching, orthoses); post-op posterior splint at 10° DF x2wks NWB then FWB + flexibility ex's Ctl: None
Engström et al. (2010)	IV Cohort study without concurrent control group; Prospective	Children with ITW with no prior treatment	*n*=15	5-13y	BoNT-A: injection to GM/ SOL + program of stretching calf muscles 5x/ wk + instructed to walk on heels at least 50 steps/day Ctl: None
van Bemmel et al. (2012)	IV Cohort study without concurrent control group; Retrospective	Children with ITW and equinus contracture	*n*=55	6-16y	Sx: percutaneous muscular GM lengthening after min 6mo non-operative treatment (serial cast, AFO, PT); post-op casting x 6wks + PT at wk 12 for 2x/ wk x 3mo Ctl: None
Engström et al. (2013)	II-M (4/7) Small RCT	Children with ITW with no prior treatment	*n*=47 26 BX 21 CA	5-15y	BoNT-A + cast: injection to GM/SOL → 4wks casting 1-2wks post-BoNT-A + program of GM stretching + instructed to walk on heels at least 50 steps/day Ctl: cast alone + ex program

Group Studies: Citations	Level of Evidence, Conduct Rating and Research Design	Participants	Total *n*	Ages	Intervention
Pistilli et al. (2014)	V Case report; Prospective	Child with persistent toe walking + frequent falls	*n*=1	18 mo	Cast: series of 4 sets BK casts over 35 days, each lasting 6-15days then AFO use with night stretch strap Ctl: None
Fanchiang Geil, Wu, Chen, & Wang (2015)	IV Cohort study without concurrent control group; Prospective	Children with ITW; Children with typical gait	*n*=30 15 ITW 15 TD	4-10y	Vibration: stood barefoot for 1 min on Soloflex whole body vibration machine vibrating at 30 Hz Ctl: No ITW control group
McMulkin et al. (2016)	IV Cohort study without concurrent control group; Retrospective & Prospective	Adolescents and young adults treated for ITW	*n*=8	12-25y	Sx: GM/SOL recession (Vulpius-type lengthening) or TAL (percutaneous or z-lengthening) Ctl: None
Szopa, Domagalska-Szopa, Gallert-Kopyto, Kiebzak, & Plinta (2016)	V Case report; Prospective	Child with severe ITW	*n*=1	5y	Cast: NDT-based PT + passive heel cord stretching 60 min 5x wk for 3wk, followed by bilat walking TICs + PT (balance + treadmill gait training) 60 min 5x wk for 3wk. Post-cast PT (equilibrium reaction, balance + treadmill gait training) 60 min 5x wk for 6wk
Williams et al. (2016)	IV Cohort study without concurrent control group; Prospective	Children with ITW without treatment in past 12 mos + able to walk heel-toe	*n*=15	4-10y	Vibration: 5 sets x 1 min vibration/1 min rest standing in supported semi-squat position on Galileo tilt table vibrating at 15 Hz; feet flat on platform, back straight against tilt table

Group Studies: Citations	**Level of Evidence, Conduct Rating and Research Design**	**Participants**	**Total *n***	**Ages**	**Intervention**
Herrin & Geil (2016)	II-W (3/7) Small RCT	Children with ITW	*n*=18	3-8y	Orthotics: custom FO + attached carbon fibre footplate during daytime (except for sports) x 6wk Ctl: Custom AFOs with PF stop during daytime (except for sports) x 6wk
Sätilä et al. (2016)	II-W (3/7) Small RCT	Children with toe walking for at least 6 mo + no contractures	*n*=29 16 BTX 13 Ctl	2-9y	BoNT-A: injections to GM-SOL + repeated at 6mo intervals to 18mo as needed + control conservative treatment Ctl: indoor shoes with firm heel cup + straps; soft cast night splints worn 5 nights/ wk; home stretch program 5x wk x 10 min/day supervised by PT 1x wk
Single Subject Design Studies: Citations	**Level of evidence, conduct rating and research design**	**Participants**	**Total *n***	**Ages**	**Intervention**
Clark et al. (2010)	III-M (9/14) Non-randomized, not-concurrent variable baseline; Prospective	Children with ITW	*n*=5	35-65mo	Baseline: multiple gait measurements collected 5-6x during this phase PT: motor control intervention 2 x 1hr sessions per wk x 9 wks; gait measures collected weekly at 2 f/u sessions

Sx, surgery; CP, cerebral palsy; ITW, idiopathic toe walking; BK, below-knee; wk(s), week(s); NWB, non-weight bearing; WB, weight bearing; Ctl, control intervention; DF, dorsiflexion; ex's, exercises; mo, months; ROM, range of movement; AFO, ankle foot orthosis; CA, cast; GM, gastrocnemius muscle; PT, Physical Therapist or Physical Therapy; TAL, tendo-Achilles lengthening; SOL, soleus muscle; re-ax, re-assessment; BoNT-A, Botulinum toxin A; AAFOs, articulated ankle foot orthosis; TA, tibialis anterior; NSAIDs, non-steroidal anti-inflammatory drugs; TD, typically developing; NDT, neurodevelopmental treatment principles; TICs, tone-inhibiting casts; FO, foot orthotics; f/u, follow-up.

Table 2.3a. Conduct of Group Design Studies for Studies with Levels of Evidence I, II, III

Study	Level/Quality	1	2	3	4	5	6	7
Sticker & Angulo (1998)	III - W (2/7)	yes	no	no	no	no	yes	no
Eastwood et al. (2000)	III - W (3/7)	yes	yes	no	no	no	yes	no
Engström et al. (2013)	II - M (4/7)	yes	yes	no	no	yes	no	yes
Herrin & Geil (2016)	II - W (3/7)	yes	yes	no	no	no	yes	no
Sätilä et al. (2016)	II - W (3/7)	yes	yes	no	no	yes	no	no

Weak (W): 1-3, Moderate (M): 4-5, Strong (S): 6-7

Conduct Questions:

1. Were inclusion and exclusion criteria of the study population well described and followed?
2. Was the intervention well described and was there adherence to the intervention assignment (for 2-group designs, was the control exposure also well described)? Both parts of the question need to be met to score 'yes'.
3. Were the measures used clearly described, valid and reliable for measuring the outcomes of interest?
4. Was the outcome assessor unaware of the intervention status of the participants (i.e., were the assessors masked)?
5. Did the authors conduct and report appropriate statistical evaluation including power calculations? Both parts of the question need to be met to score 'yes'.
6. Were dropout/loss to follow-up reported and less than 20%? For 2-group designs, was dropout balanced?
7. Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?

Table 2.3b.
Conduct of Single Subject Design Studies for Studies with Levels of Evidence I-III

Study	Level/ Quality	1	2	3	4	5	6	7	8	9	10	11	12	13	14
Clark et al. (2010)	III - M (9/14)	y	y	y	y	n	n	n	y	n	n	y	y	y	y

Weak (W): 1-6, Moderate (M): 7-10, Strong (S): 11-14

Conduct Questions:

1. Was/were the participant(s) sufficiently well described to allow comparison with other studies or with the reader's own patient population?
2. Were the independent variables operationally defined to allow replication?
3. Were intervention conditions operationally defined to allow replication?
4. Were the dependent variables operationally defined as dependent measures?
5. Was inter-rater or intra-rater reliability of the dependent measures assessed before and during each phase of the study?
6. Was the outcome assessor unaware of the phase of the study (intervention vs. control) in which the participant was involved?
7. Was stability of the data demonstrated in baseline, namely lack of variability or a trend opposite to the direction one would expect after application of the intervention?
8. Was the type of SSRD clearly and correctly stated, for example, A-B, multiple baselines across subjects?
9. Were there an adequate number of data points in each phase (minimum of five) for each participant?
10. Were the effects of the intervention replicated across three or more subjects?
11. Did the authors conduct and report appropriate visual analysis, for example, level, trend and variability?
12. Did the graphs used for visual analysis follow standard conventions, for example, x- and y-axes labeled clearly and logically, phases clearly labeled (A, B, etc.) and delineated with vertical lines, data paths separated between phases, consistency of scales?
13. Did the authors report tests of statistical analysis, for example, celebration line approach, two-standard deviation band method, C-statistic, or other?
14. Were all the criteria met for the statistical analyses used?

Table 2.3c. Conduct of Systematic Review Studies

Study	**Level/Quality**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**
van Bemmel et al. (2014)	II - M	y	y	y	y	n	n	y	y	y	M
van Kuijk et al. (2014)	II - M	y	y	y	y	n	n	n/a	n/a	y	M

M, moderate

Conduct Questions:

1. Were the search methods reported?
2. Was the search comprehensive?
3. Were the inclusion criteria reported?
4. Was selection bias avoided?
5. Were the validity criteria reported?
6. Was validity assessed properly?

7. Were the methods used to combine studies reported?
8. Were the findings combined appropriately?
9. Were the conclusions supported by the reported data?
10. What was the overall scientific quality of the overview?

2.4.5 Outcomes, Measures and Results

Results from all studies were coded for ICF components by the intervention's outcome of interest (World Health Organization, 2007). This review identified 12 treatment outcomes for children with ITW: one was coded at the body structures level (muscle-tendon length); five were coded at the body functions level (gait pattern, ankle ROM, ankle strength, vibration perception, pain); two were coded at the activity level (gross motor development, lower extremity function/mobility); two were coded at the participation level (keeping up with peers, sporting activities); one was aimed at the environmental level (parental satisfaction); and one was aimed at the personal level (subject satisfaction). All 28 intervention studies included in the review described a motor outcome as one of their primary outcomes. Two studies documented gross motor development (Clark et al., 2010; Pistilli et al., 2014) and four other studies reported on mobility or lower extremity motor activities (Jacks et al., 2004; McMulkin et al., 2016; Herrin & Geil, 2016; Sätilä et al., 2016). One study used a telephone survey to assess whether or not children with ITW were able to keep up with their peers following treatment (Kogan & Smith, 2001) and another study used a questionnaire to determine the subjects' perceived limitation in sporting ability or level of walking (Stott et al., 2004). Three studies assessed parental satisfaction of their child's outcome using study-specific parent survey or questionnaires (van Bemmel et al., 2014; Kogan & Smith, 2001; Herrin & Geil, 2016); only two studies assessed the subjects' own satisfaction with their treatment outcome (Stott et al., 2004; McMulkin et al., 2016).

Table 2.4 summarizes the highest level of evidence for ITW interventions and is limited to studies rated as levels I to III. One of the intervention studies with the highest level of evidence (level II - M) compared casting alone and casting with BoNT-A, using treatment outcomes all targeted at the body function level of the ICF, and demonstrated that the addition of BoNT-A before casting did not improve outcomes with respect to gait, ankle DF PROM or ankle DF strength (Engström et al., 2013). Another RCT (II-W) compared conservative treatment alone and conservative treatment in conjunction with repeated BoNT-A injections; however, treatment outcomes were aimed at the activity level as well as the body function level (Sätilä et al., 2016). They also found the addition of BoNT-A did not improve toe walking, ankle DF PROM or lower extremity function (Sätilä et al., 2016). The only other RCT (II-W) compared an articulated ankle foot orthosis (AFO) with custom foot orthotic (FO), utilizing treatment outcomes targeted at the body function, activity, and environmental levels of the ICF (Herrin & Geil, 2016). The AFOs diminished the effects of toe walking more than the FOs when the orthoses were worn; however, once the orthoses were removed and gait was re-assessed at follow-up, only the group previously wearing FOs demonstrated significant improvement (Herrin & Geil, 2016). No significant differences in functional mobility or parental satisfaction between groups were found (Herrin & Geil, 2016).

Two of the studies with lower levels of evidence (III-W) compared casting and surgery with a control group – again, with outcomes addressing the body function component of the ICF (Eastwood et al., 2000; Stricker & Angulo, 1998). The control groups underwent observation alone (Eastwood et al., 2000) or were either observed, monitored for footwear changes, or given stretching exercises (Stricker & Angulo, 1998). One of the studies in the evidence table evaluating an outcome beyond the component of body functions examined a motor control

intervention twice weekly for nine weeks (Clark et al., 2010). The primary outcomes were gross motor development and ankle DF PROM, targeting activity and body function components, respectively, yet the motor control intervention failed to demonstrate a shift to heel-toe gait.

The predominant focus on body functions is reflected further in the systematic review by van Bemmel and colleagues (2014). They compared 10 studies, between 1998 and 2012, included in this review, using casting or surgical treatment to target body functions (ankle DF, toe walking). Only three studies in their review considered parental satisfaction of the treatment and associated outcome (van Bemmel et al., 2014). Although parental satisfaction was reported in four of the studies they reviewed, only three truly considered satisfaction at a contextual level as the nature of one of the study-specific parent questionnaires was directed at post-treatment gait patterns rather than to the level of parental contentment. The authors did not pool these outcomes for statistical analysis given the variability of actual measurements used. The weighted means of passive ankle DF were calculated by Mann-Whitney *U* test and toe walking means were calculated by chi-square test. van Bemmel et al. (2014) concluded that surgery is superior to casting for improving dorsiflexion PROM yet there was no significant difference in the persistence of toe walking between groups. A more recent systematic review extended this opinion by adding that while surgical intervention appears to result in better improvement of ankle DF PROM than casting; only studies addressing the effects of surgery are able to show effects lasting more than one year (van Kuijk et al., 2014).

Table 2.4.
Summary of Studies: Outcomes, Measures, and Results (Levels of Evidence I-III)

Group Studies: Citations	Outcome of Interest	Measure	Body Structure/s Body Functions	Activities and Participation	Contextual Factors
Stricker & Angulo III-W (1998)	Ankle DF PROM Parental satisfaction of time child spent TW	Goniometry Questionnaire	Sx: ss↑DF Sx: p<0.05		
Eastwood et al. III-W (2000)	Toe Walking	Linear analog scale	ss ↓TW p=0.0001 for all groups; ns difference b/w control + cast group		
Engström et al. II-M (2013)	Toe Walking	3DGA	ns diff b/w groups		
	ITW Severity	Alvarez et al. (2007)	ns diff b/w groups		
	Ankle DF PROM	Goniometry	ns diff b/w groups		
	Ankle DF strength	Dynamometry	ns diff b/w groups		
	Parent perception of TW frequency	Questionnaire	ns diff b/w groups		
Herrin & Geil II-W (2016)	Temporal spatial characteristics	3DGA	ss ↑ velocity wearing AFO p=0.006		
	Heel rise timing	3DGA	ss imp timing in AFO + FO p<0.001; ss imp timing FO removed p<0.001		
	Ankle DF PROM	Not reported	ns diff b/w groups		
	Parent perception of TW frequency	Survey	ns diff b/w groups		

Group Studies: Citations	Outcome of Interest	Measure	Body Structure/s Body Functions	Activities and Participation	Contextual Factors
Herrin & Geil (2016)	LE functional mobility	L Test		ns diff b/w groups	
	Parent satisfaction	Survey			ns diff b/w groups
Satila et al. II-W (2016)	Toe walking	Video analysis; TWSS	ns diff b/w groups		
	Ankle DF PROM	Goniometry	ns diff b/w groups		
	LE function	LEFS		ns diff b/w groups	
Single Subject Design Studies: Citations	**Outcome of Interest**	**Measure**	**Body Structure/s Body Functions**	**Activities and Participation**	**Contextual Factors**
Clark et al. III-M (2010)	Gross motor development	GMQ-PDMS-2		p=0.022 for group GMQ comparisons but ns post-hoc comparisons	
	Ankle DF PROM	Goniometry	p=0.002		
	Toe Walking	Parent VAS; GED	ns		

Sx, surgery group; ss, statistically significant; p, significance level; TW, toe walking; ns, not statistically significant; b/w, between; 3DGA, 3-dimensional gait analysis; imp, improved; LE, lower extremity; L-test, L Test of Functional Mobility; TWSS, Toe Walking Severity Scale; LEFS, Lower Extremity Functional Scale; PDMS-2, Peabody Developmental Motor Scales - Revised; GMQ, gross motor quotient; VAS, visual analog scale; GED, gait event detector.

2.4.6 Evidence for Intervention Effects on Outcomes Representing Body Functions

Toe walking, in the body function component, was the only outcome of interest described in all six level I-III studies included in the evidence table (Table 2.4). Three of the six studies evaluated the effect of serial casting on toe walking with a minimum of four weeks of casting

across all groups (Eastwood et al., 2000; Stricker & Angulo, 1998; Engström et al., 2013). Two of the studies demonstrated a significant reduction of toe walking; these groups compared casting with observation (no treatment), surgery, or the addition of BoNT-A to casting (Stricker & Angulo, 1998; Engström et al., 2013). Conversely, the study evaluating casting or use of orthotics as 'casting' failed to produce a significant difference (Eastwood et al., 2000). One concern with these findings from the latter study stems from the heterogeneity of the treatment approach: protocols of either six to 12 weeks in below-knee casts or three to eight months in solid ankle foot orthotics (AFOs) are grouped into the casting comparison. The duration of time spent in AFOs is unknown but it is unlikely they were worn all day as is typically the case with below-knee casting. This study also included an observational control group and found no significant difference in toe walking between the control and cast groups (Eastwood et al., 2000). The study comparing casting alone and casting with BoNT-A, measured toe walking by three-dimensional gait analysis (kinematic and kinetic data) and classified ITW severity; the authors found significant improvements in gait parameters and level of severity in both intervention groups (Engström et al., 2013). Compared to previous small sample studies addressing the use of BoNT-A with ITW (Jacks et al., 2004; Brunt et al., 2004; Engström et al., 2010), no significant difference was found between the BoNT-A and cast group and the cast alone group, although 81% of the children continued to toe walk following treatment (Engström et al., 2013). Another study also considered toe walking severity but compared the effects of BoNT-A to a conservative treatment regime with similar results; both treatment groups demonstrated improved severity with no between group differences found, although the BoNT-A group attained improvements earlier (Sätilä et al., 2016). This study developed a different non-validated severity scale, had a younger and smaller sample size, used night splints instead of casting following injections, and

used higher doses of BoNT-A, making comparison difficult. BoNT-A injections were also repeated systematically, on an as-needed individual basis, increasing the heterogeneity within that treatment group.

The two studies which evaluated the effect of surgery with either tendo-Achilles lengthening (TAL) (Eastwood et al., 2000; Stricker & Angulo, 1998) or gastrocnemius recession (Stricker & Angulo, 1998) found a significant improvement in toe walking, although complete resolution did not occur in either study. Children receiving surgery were generally older and demonstrated more ankle DF restrictions than children in conservative treatment groups. Using a conservative treatment approach, when children were treated with a motor control intervention, no significant change in toe walking was observed during daily life as measured by a gait event detector and parent report (Clark et al., 2010). However, study limitations, such as a small sample size, short intervention period, and lack of monitoring of home program adherence, likely contributed to these results. Another conservative treatment approach was used in a study comparing the effects of two distinct orthoses on timing of heel rise as a function of toe walking (Herrin & Geil, 2016). Although timing of heel rise improved in both groups when wearing orthotics, only the FO group showed improved carry-over results when wearing shoes without orthotics. This study was also limited by a short intervention period and it was unclear whether any other treatment was received or what individual activity levels were during the intervention phase. Several studies in this current review examined parental satisfaction or perception of toe walking, measured with a study-specific questionnaire, survey, or scale (Fox et al., 2006; Eastwood et al., 2000; Stricker & Angulo, 1998; Stott et al., 2004; Clark et al., 2010; Hirsch & Wagner, 2004; Kogan & Smith, 2001; van Bemmel et al., 2012; Herrin & Geil, 2016). Though a critical outcome to consider, the questionnaires used to evaluate parental report were not

standardized. Further, in some studies, it is questioned whether reporting is based on satisfaction with toe walking alone or based on the impact of toe walking in daily life (Stricker & Angulo, 1998; Stott et al., 2004; Hirsch & Wagner, 2004; Kogan & Smith, 2001; van Bemmel et al., 2012, Herrin & Geil, 2016).

When examining the body function component, ankle DF PROM was used to measure change in four of the six studies included in the evidence table (Stricker & Angulo, 1998; Clark et al., 2010; Engström et al., 2013, Sätilä et al., 2016). Three studies showed significant improvement in ankle DF PROM, although one study found significant improvement in the surgical group only (Stricker & Angulo, 1998); in the cast alone compared to cast and BoNT-A study, no significant difference was found between groups (Engström et al., 2013). One study only found improved passive ankle DF in 52% of participants (Sätilä et al., 2016). Goniometry was used in all four studies to measure ankle DF PROM. Ankle strength, measured by dynamometry, was the only other body function targeted by the interventions in the included studies; Engström and colleagues found a significant increase in strength in both 'cast alone' and 'BoNT-A' groups but with no significant differences between groups (Engström et al., 2013).

2.4.7 Evidence for Intervention Effects on Outcomes Representing Activities

Gross motor development and lower extremity function/functional mobility were the only outcome measures in the evidence table targeting the ICF-CY component of activity. However, while children receiving a motor control intervention experienced improved gross motor skills, assessed by the Peabody Developmental Motor Scales - Revised (PDMS-2) (Fewell & Folio, 2000), these failed to reach statistical significance in post-hoc comparisons (Clark et al., 2010). It is possible that ITW in children four years and older is not a result of motor control deficiency - the premise of this intervention. Other possibilities remain that the lack of a significant change

in gait pattern may be a result of the variability seen in outdoor ambulation, a treatment protocol which was shorter and less intense than previous recommendation (Conrad & Bleck, 1980), or a lack of flexibility in addressing individual needs. Sätilä et al. (2016) asked parents to determine their child's level of difficulty with a number of lower extremity functional activities before and after conservative treatment with or without BoNT-A and night splints. This evaluation was included as an activity measure but is intended as a self-report questionnaire for adults, resulting in decreased confidence in results. The only other reference to the assessment of activity limitations is by Herrin and Geil (2016). They used a test of basic mobility skills at baseline and follow-up with no between group differences found, yet description of the test and how it was performed was lacking.

2.4.8 Evidence for Intervention Effects on Outcomes Representing Other Components of the ICF

Parental satisfaction was the sole outcome measure in the evidence table aimed at the ICF-CY component of environmental factors. No description of the parent survey was included in the single study (Herrin & Geil, 2016); however, it appeared to assess for parent satisfaction of gait following treatment as well as the parents' opinion on the effectiveness of orthotic treatment. No significant between group differences were found for either component of the survey, yet parents reported 63% improvement in gait with the AFO compared to 38% improvement with the FO; in contrast, parents considered the AFO to be ineffective in 50% of the participants whereas the FO was rated as 56% effective. Unfortunately, without a validated survey or an understanding of the components of the survey, it is difficult to interpret these results.

No other interventions were found in the level I-III studies targeting the participation, environment, or personal factors components of the ICF-CY; thus, evidence findings from the

level IV-V studies (Table 2.2) will contribute to this discussion. These results either lacked statistical significance, valid measurements, or study rigor; nevertheless, information from these studies may inform future directions for clinical practice. A follow-up parental telephone survey conducted from three months to 6.5 years postoperative evaluation of 10 out of 15 children who had undergone a percutaneous TAL demonstrated that all 10 children were able to keep up with their peers (Kogan & Smith, 2001). Similarly, a questionnaire conducted with 13 subjects 5.4 to 15.6 years post-serial casting, TAL, or gastrocnemius recession, who were now 16 to 25 years old, found that only one subject experienced minor difficulties with sporting activities (Stott et al., 2004). Furthermore, this questionnaire showed that all subjects were satisfied with the treatment given and that 12/13 subjects would choose to undergo the same treatment (Stott et al., 2004). One other study conducted a descriptive interview with all eight participants in a similar age range, 5.1 to 15.5 years post-surgical lengthening for gastrocnemius/soleus contractures; self-report revealed all participants were satisfied with their surgical outcome, there were no mobility problems or activity restrictions (although two participants reported mild pain with longer activity duration), and only one participant had a problem with tripping and falling (McMulkin et al., 2016). These two studies attempted to address the personal factors level. Telephone surveys were used in two other studies to determine parental satisfaction with treatment outcomes which addressed the environmental factors level of the ICF (Kogan & Smith, 2001; van Bemmel et al., 2012). One study reported that all parents contacted for follow-up (10/15 children) were satisfied with their child's outcome, while another found that parents scored between four to 10 on a 10-point visual analogue scale with a mean satisfaction rating of 8/10 (van Bemmel et al., 2012). No evidence was found for associations between different components of the ICF in any of the studies included for review.

2.4.9 Adverse Events of Treatment Approaches for ITW

Four group research design studies, and one single subject research design study with level II to III evidence, documented adverse events in 284 children (97 observation only, 79 casting/orthotics, 61 surgery, 42 casting and/or BoNT-A, five therapy activities) (Table 2.5). Seven children experienced complications following cast treatment including pressure ulcerations (Stricker & Angulo, 1998), mid-calf pain, itching, and chafing (Engstrom et al., 2013). Only one complication after surgical treatment resulted in increased severity of toe walking based on parental report; although the physician reported a normal gait pattern and no evidence of over lengthening the Achilles tendon (Eastwood et al., 2000). The cast and BoNT-A group reported seven complications including post-injection calf pain, mild calf pain during casting, and minor skin problems (Eastwood et al., 2000). The conservative treatment and BoNT-A study had 38 adverse events reported by parents including calf tenderness, clumsiness, irritability, fatigue, muscle weakness, appetite loss, flu-like symptoms, constipation, urgency, and a rash (Sätilä et al., 2016). One child reported one incidence of leg pain associated with therapy activities during a motor control intervention phase (Clark et al., 2010). Of these reports, four out of five studies evaluated complications by parental report (Eastwood et al., 2000; Clark et al., 2010; Engstrom et al., 2013; Sätilä et al., 2016) and one failed to report the documentation system (Stricker & Angulo, 1998). All of the adverse events were reported in sufficient detail although follow-up evaluation ranged from one to 22 years and parent questionnaires were study specific. One study did not state the method used to determine adverse events (Stricker & Angulo, 1998).

Additionally, five group research design studies with level IV evidence reported adverse events in 162 children (seven surgery, one casting) (Table 2.5). These studies are less rigorous

and the information extracted must be approached with caution. Seven reported adverse events were described in the surgical group including Achilles tendinitis (Kogan & Smith, 2001; Hemo et al., 2006), excessive ankle DF (Jahn et al., 2009), wound dehiscence (Hemo et al., 2006), and intraoperative ankle fracture (van Bemmel et al., 2012). Of the 44 children treated with serial casts, there was only one report of plaster sores caused by the end of the cast (Fox et al., 2006). Three of these studies failed to report their method for collecting data on complications (Fox et al., 2006; Hemo et al., 2006; Jahn et al., 2009), one was reported by parental telephone survey conducted 3.5 to six months post-operatively (Kogan & Smith, 2001), and one was a post-operative surgeon's report (van Bemmel et al., 2012). Further, one case report with level V evidence reported pressure areas and skin irritation following the application of tone-inhibiting casts for three weeks (Szopa et al., 2016).

Table 2.5. Reported Adverse Events

Group Studies: Citations	**Level of Evidence**	**Total *n***	**Method of Ascertaining Adverse Events**	**Description of Adverse Events Reported**
Stricker & Angulo (1998)	III	80	Not reported	Cast/AFO group: partial- thickness skin pressure ulcerations on dorsum of feet (*n*=2)
Eastwood et al. (2000)	III	136	Parent-determined assessments of TW severity 2-22 years after presentation	Parents believed child was worse (authors state no evidence of over-lengthening Achilles tendon + gait normal according to physician-determined outcome (*n*=1)
Kogan & Smith (2001)	IV	10	Parent QNR 3 mo-6.5 years post-op	Occasional Achilles tendinitis post-op (*n*=2)
Hemo et al. (2006)	IV	15	Not reported	Immediate post-op superficial wound dehiscence (*n*=1) + Achilles tendinitis 6 years post-op (*n*=1)
Fox et al. (2006)	IV	44	Not reported	Plaster sores caused by proximal end of cast (*n*=1)
Jahn et al. (2009)	IV	38	Not reported	Excessive ankle DF post-op (*n*=2)

Group Studies: Citations	Level of Evidence	Total *n*	Method of Ascertaining Adverse Events	Description of Adverse Events Reported
van Bemmel et al. (2012)	IV	55	Surgeon report	Ankle fracture during surgery resulting in required open reduction (*n*=1)
Engström et al. (2013)	II	47	Parent report	Cast group: mid-calf pain during casting (*n*=2) + itching and chafing from casts (*n*=3) Cast + BX: calf pain post BX injection (*n*=1), mild calf pain during casting (*n*=3) + minor skin problems (*n*=3)
Szopa et al. (2016)	V	1	Not reported	Pressure areas + skin irritation post-casting (*n*=1)
Sätilä et al. (2016)	II	29	Open parent QNR	BX group: calf tenderness (*n*=13); clumsiness (*n*=9); irritability (*n*=3); fatigue (*n*=5); m/s weakness (*n*=3); appetite loss (*n*=1); flu-like symptoms (*n*=1); constipation (*n*=1); urgency (*n*=1); rash (*n*=1)
Single Subject Design Studies: Citations	**Level of Evidence**	**Total *n***	**Method of Ascertaining Adverse Event**	**Description of Adverse Events Reported**
Clark et al. (2010)	III	5	Parent report	Leg pain associated with therapy activities (*n*=1); therapist observed small lower leg contusions possibly due to bumping into furnishings

AFO, ankle foot orthosis; TW, toe walking; QNR, questionnaire; mo, months; post-op, post-operatively; DF, passive ankle dorsiflexion range of motion; BX, Botulinum toxin A; m/s, muscle

2.4.10 Strength of the Evidence for Treatment Strategies

Evidence for ITW interventions has been summarized based on six of the 28 intervention studies included in this review and two systematic reviews assigned ratings of levels I to III. Two of the studies and the two systematic reviews were rated as moderate in quality and four of the

studies were found to be weak in quality (Tables 2.3a, b, c). Methodological limitations of group design studies included: (1) lack of valid and reliable outcome measures, including questionnaires for parental satisfaction; (2) lack of blinding of assessors to participant intervention status; (3) imbalanced drop-out to follow-up rate in two group design; (4) inadequate reporting of statistical evaluation, including power calculations; and (5) lack of control groups or insufficient description of control exposure to allow replication. The results of ITW treatment effects in group design studies are based on a small sample of 300 children and only 47 of these children were represented in a recent study of moderate quality (Engström et al., 2013). Methodological weaknesses of the single subject design study (Clark et al., 2010) included are: (1) lack of baseline stability demonstrating variability for the outcome measuring heel strike frequency; (2) insufficient level of data points in the intervention phase for each participant (n=5) with a majority of insignificant upward trends for most participants and no change for one participant; (3) lack of reliability of the dependent measures, including measures of ankle PROM with goniometry; (4) failure to replicate the effects of the intervention over a sufficient number of participants; and (5) lack of a representative sample of children presenting for ITW interventions by physical therapy. Given the very small sample size and lack of significant findings in the single subject design study, it is difficult to inform clinical practice based on their results.

2.4.11 Summary of Literature Review and Study Aims

Through review of the literature, preliminary evidence exists to suggest that serial casting and surgery have a short term effect of increasing ankle DF PROM at the body functions level. No connection was found between ankle DF PROM and persistent toe walking at the body

function level of the ICF-CY. No effective interventions were found at any other levels of the levels of activities, participation, environment, or personal factors within the ICF framework.

The evidence to support the efficacy of treatment approaches for ITW is limited by a large number of retrospective studies, a lack of reliable measurements used consistently across studies, and studies lacking internal validity. In short, we do not know if children with ITW are affected functionally and if there are truly long-term consequences. Researchers and clinicians together need to consider appropriate methods for accurate assessment of ITW given the lack of correlation between physical evaluation and gait analysis. Goal setting with children and their family is essential in the management of ITW; particularly since it is unlikely a connection exists between passive ankle DF angle and normalized gait.

At the Shriners Gait Laboratory at Sunny Hill Health Centre for Children (SHHCC), we developed an ITW classification framework based on a cohort of 133 children with ITW, recruited from 1997 to 2005 (Alvarez et al., 2007). Severe toe walkers consistently demonstrated limited ankle DF PROM, limited ankle movement during gait, and abnormal ankle rocker formation leading to recommended treatment for these children (Alvarez et al., 2007). Moderate toe walkers were recommended treatment less frequently; only as requested by families or if complaints persisted.

As a result of limited conclusive research describing the natural history of ITW and efficacy of treatment approaches, a follow-up research project of the children in our original classification study (Alvarez et al., 2007) was conducted with the following three objectives: 1) To describe the natural history of ITW and the effect of treatment on long-term outcomes of body functions and structures in adolescents and young adults; 2) To determine the relationships

between ITW severity and treatment on long-term outcomes of body functions and structures; 3) To explore activity limitations and participation restrictions in these adolescents and young adults.

Hypotheses: 1) Children assessed for ITW in our Gait Lab would demonstrate improved gait parameters as adolescents and young adults irrespective of whether or not treatment was received; 2) Children classified at increasing levels of severity would continue to demonstrate signs of abnormal gait as adolescents and young adults regardless of intervention received; 3) Children and young adults with ITW would demonstrate minimal limitations in activity or restrictions in participation in daily living.

3. METHODS

3.1 Design and Rationale

A retrospective cohort study with concurrent control group was used to meet the purpose and objectives of this project. At present there is limited research exploring long-term outcomes of ITW into adolescence and adulthood; therefore, this follow-up study provided a descriptive, explorative, and analytical approach to evaluating change over time in this population sample (Portney & Watkins, 2009). The non-experimental design was used to describe the natural history of ITW and to analyze the relationships between time, treatment, and severity to current status as reflected by long-term gait outcomes. This strategy allowed for the exploration of how current gait status had changed since the initial assessment to generate hypotheses about the need for conservative or non-conservative interventions for ITW, testable in future studies using experimental designs (Portney & Watkins, 2009). No additional intervention was provided in this study which examined the natural history, post-intervention phase, and capacity for activities and participation in this cohort.

3.2 Participants

3.2.1 Ethics and Recruitment

Ethics approval for this project was obtained through the University of British Columbia Children's and Women's Research Ethics Board. All recruitment was conducted by the master's candidate and occurred through the Shriners Gait Lab (SGL) at SHHCC in Vancouver. Purposive sampling was used to follow-up with the original cohort of 133 children based on their inclusion in our primary classification study from 1997 to 2005 (Alvarez et al., 2007). Families were contacted by mail via their most recent address in electronic medical records and informed

of the proposed follow-up study. All potential participants received an information letter (Appendix A), consent (Appendix B) and assent forms (Appendix C). Families and/or participants were given a two-week period to review the information letter and consent forms prior to being contacted by telephone to provide additional information about the project, obtain informed consent, and schedule a follow-up gait lab assessment for consenting participants.

3.2.2 Participant Inclusion and Exclusion Criteria

Recruitment of our original sample of 133 children was based on the following inclusion criteria: referral by a pediatric orthopaedic surgeon with a diagnosis of idiopathic toe walking and between four to 16 years of age. To ensure the sample was exclusively children with ITW, physical examinations and neurological assessments were reviewed prior to gait analysis by the study physical therapist and an orthopedic surgeon (Alvarez et al., 2007). Post hoc analysis of collected data was performed during gait data analysis to further screen for any other potential ankle pathology and no children were excluded at that point. The children who participated in the original study were between four and 16 years of age (mean 8.8 years) and consisted of 65 females and 68 males.

To observe change at follow-up and to determine statistical significance, a sample size target of 44 adolescents and/or young adults whose data was used in the original ITW classification study was estimated for the current study. A sample size calculation was done using G*Power (Buchner, Erdfelder, Faul, & Lang, 2009; Faul, Erdfelder, Land, & Buchner, 2007) and a recent RCT that classified ITW severity, measured ankle DF PROM, and compared the use of casting with and without BoNT-A (Engström et al., 2013). It was based on their data of ankle angle measurements during gait, reported effect size of 0.50, alpha error probability of

0.05, and power of 0.80. As this study design is not a RCT, the participant size represents a sample of convenience that may help guide future study sample estimation.

Adolescents and young adults from the original study were excluded from the follow-up study if they now had a diagnosis of cerebral palsy, spinal cord abnormality, myopathy, peripheral neuropathy, neuromuscular disorder, movement disorder, autistic spectrum disorder, talipes equinovarus, unilateral toe walking, sudden onset toe walking, or any other diagnosis that affected their range of motion at the ankle.

3.2.3 Participant Sample

A total of 45 adolescents and young adults met both the inclusion and exclusion criteria and were recruited for this study. Although they all completed follow-up clinical gait lab assessments, one young adult was removed from the data set due to an incomplete baseline assessment in 2004. One participant had recently received a 10-year follow-up assessment in the SGL by the master's candidate as part of routine clinical follow-up; therefore, gait lab data was included retrospectively and questionnaire data included prospectively once informed consent was received. Of the original 133 children, it was not possible to contact 48 of them, 33 declined and seven were not applicable due to differential diagnoses including cerebellar atrophy, vascular malformation, peripheral neuropathy, autism, severe hemophilia A, juvenile rheumatoid arthritis, and one was deceased. The declining potential participants were too busy, unable to travel, could not afford the cost of travel or did not provide a reason. The remaining 44 adolescents and young adults had complete data sets and were included in all study analyses.

Data analyses were completed based on whether or not participants reported receiving treatment. Twenty participants reported not receiving any treatment following their initial assessment other than recommendations for incorporating stretching exercises into their daily

routine to maintain ankle DF ROM. This group was designated as the non-treatment group for concurrent control. Of these 20 participants, three had been given foot orthotics and seven had undergone serial casting followed by AFOs in their community *prior* to their initial gait lab assessment. Twenty-four participants reported receiving treatment following their initial assessment; all but four of them received treatment within the same year, two received treatment within two to three years following and two received treatment six years' post-initial assessment with mean follow-up after intervention of 11.8 years (standard deviation, SD, 3.4). This group was considered the treatment group for comparison. Six participants received serial casting for six weeks with a change in cast after three weeks and 17 participants received BoNT-A injections, prior to serial casting for six weeks; all 23 participants wore AFOs for one year following cast removal. Participants were treated by one of two pediatric orthopaedic surgeons. No participants reported a lack of adherence to intervention. Participant descriptive information is outlined by treatment group in the Results Chapter (Table 4.1).

Participants receiving BoNT-A injections were not differentiated from those only receiving casts and AFOs as current evidence demonstrates no significant difference in gait parameters, severity, or ankle DF PROM between casting following BoNT-A or cast-alone groups (Engström et al., 2013). Only one participant was treated with bilateral percutaneous TAL and was also included in the treatment group without differentiation as the systematic reviews by van Bemmel et al. (2014) and van Kuijk et al. (2014) show no significant differences between casting and surgical groups with respect to persistent toe walking. The only difference reported in the included studies was an increase in ankle DF PROM in the surgical group compared to the cast group (van Bemmel et al., 2014) posttreatment. No significant difference was found in the one participant in our study treated with TAL ($p=0.822$, 95% Confidence Interval, CI, [-1.1, 0.9],

p=0.660, 95% CI [-1.3, 0.8] for left and right sides, respectively). The ankle kinematics and kinetics during gait in this participant were all within the range of values found in the other treated participants. No participants were receiving treatment at the time of follow-up visit.

3.3 Outcome Measures

Several outcome measures were collected in this study to explore how current gait status and ITW severity had changed since the initial assessment as well as to determine if there were any activity limitations or participation restrictions in these adolescents and young adults. Demographic information was collected for each participant, including age (years), height (cm), body mass (kg) and sex, and are outlined in the Results Chapter (Table 4.1). Three dimensional computerized gait data and physical exam measurements were collected systematically at follow-up to repeat the gait analysis procedures used in the initial study. Quantitative gait data were dynamic and representative of locomotor function whereas the physical exam, in particular ankle DF PROM, was static and measured prior to locomotion. Additionally, the third outcome measure was administered at the follow-up visit only to assess current functioning and disability.

3.3.1 Gait Analysis Data

Clinical instrumented gait analysis studies locomotion and determines what causes a child, adolescent or adult to walk in a particular way. A gait cycle, or stride, occurs with the sequence of one foot contacting the ground through stance (60%) and swing (40%) until that foot contacts the ground again. Further, each gait cycle may be considered representative of how an individual walks and variability of that walking pattern can be determined by analyzing several cycles (Baker, 2013). The dynamic activity of gait can be divided into kinematics and kinetics, with kinematics describing motion and joint angles are calculated; kinetics describing the effects of forces and moments on the motion of the human body (Schwartz, 2004; Baker, 2013). The

primary outcome measure for this study was maximum ankle DF angle in stance, addressing the ICF-CY level of body function. Diminished ankle DF during stance is one of the most frequently reported gait deviations in the ITW literature. Further, justification for the study sample size was based on identifying a 5° difference in ankle angle (SD 9.5°) during gait (Engström et al., 2013). Additional data from the gait analysis were collected to support the primary outcome measure including ankle angle at initial contact, ankle DF in swing, knee extension in stance, knee flexion in swing, anterior pelvic tilt, foot orientation ankle kinetics, and electromyography (EMG) data.

The primary measures used to determine ITW severity types in the preliminary study (Alvarez et al., 2007) were identified based on previous work considering gait adaptations of the contra-lateral lower limb in children with cerebral palsy, specifically spastic hemiplegia (Sawatzky, Alvarez, Beauchamp, & Black, 1999). They included the presence or absence of a first rocker, an early third rocker (premature transition from DF to PF), and an early predominant internal ankle moment. This early predominant ankle moment is calculated from kinematic and kinetic data and reflects a plantarflexor moment in early stance that is larger than the plantarflexor moment typically present in late stance. Alvarez et al. (2007) defines the first ankle moment (AM1) to differentiate the peak PF moment during initial stance from the second, typical, peak PF moment during late stance (AM2). This has also been described as a "double bump" ankle PF moment pattern (Stott et al., 2004; Hemo et al., 2006), indicative of an early increased PF moment. Type 1 (mild) toe walking is distinguished by the presence of an ankle angle greater than -5° DF at initial contact with increasing PF (first rocker), typical third rocker timing, and predominant late stance PF moment. Type 2 (moderate) toe walking may or may not have a first rocker and typical third rocker timing and has a predominant late stance PF moment.

Type 3 (severe) toe walking has an absent first rocker, premature transition from DF to PF, and early predominant ankle moment.

The reliability of instrumented clinical gait analysis is based on regular evaluation of the measurement system performance and data collection techniques, concurrent videotaping, interdisciplinary team education and data interpretation, and accurate marker placement (McGinley , Baker, Wolfe, & Morris, 2009; Davis, 2008). This study used a conventional marker set and modified multi-segment foot model that has undergone test-retest reliability trials to determine intra-rater repeatability and inter-rater repeatability for the two physical therapists in the SGL; reliability was estimated based on mean, standard deviation (SD), and range (in degrees) (Maurer et al., 2013). One of the physical therapists performed all PT assessments, marker, and EMG placement in the preliminary study and the other physical therapist, the master's candidate, completed all physical examination, marker and EMG placement in the current follow-up study.

For the purpose of this study, as recommended by McGinley et al. (2009), repeat test-retest reliability trials were conducted prior to data collection at follow-up to report the standard error of measurement (SEM), or within-subject standard deviation, as a measure of absolute reliability (Bland & Altman, 1996) for the primary kinematic outcome variable (in degrees). The intraclass correlation coefficient (ICC) was also calculated as an indication of relative reliability (Shrout & Fleiss, 1979; Birmingham, Hunt, Jones, Jenkyn, & Giffin, 2007), although ICCs are limited due to their heavy dependence on the variation within the population and their suitability for larger sample sizes. Reliability trials were repeated with both SGL physical therapists and the results for sagittal plane ankle angles are included in Appendix D. As all gait analyses in this

study were conducted by the master's candidate, the mean, SD, SEM and ICC are reported for this candidate for the primary outcome measure of maximum ankle DF angle in stance.

Three healthy participants aged 8-41 years (average age 26 years, 3 females) were tested over three separate sessions with visits spaced between one week and two months apart. Data was collected as described in Chapter 3.4 (Procedure). Maximum ankle DF in stance for each participant was compared between testing sessions on the left side to be consistent with our preceding test-retest repeatability trials (Maurer et al., 2013) and to avoid possible bias or incorrect statistical conclusions as a result of including paired, non-independent, data from both limbs (Bryant, Havey, Roberts, & Guyatt, 2006; Sangeux, Wolfe, & Graham, 2013).The mean maximum ankle DF angle in stance for all three participants was 9.1° $\pm$ 1.4° SD with a SEM of 0.9° and an ICC (3,1) of 0.80. A SEM value of two degrees or less is considered acceptable in terms of kinematic measurement and gait data interpretation (McGinley et al., 2009).

3.3.2 Clinical Range of Motion

Clinical passive ankle DF ROM is frequently reported as an outcome measure at the body function level of the ICF-CY, particularly in earlier ITW studies addressing conservative and soft tissue surgical treatment approaches (van Kuijk et al., 2014). Maximum ankle DF PROM is a primary measurement used for treatment recommendations when three-dimensional gait analysis is not available or warranted, despite the widespread reports of a lack of correlation with ankle DF during gait and its unlikely ability to solely distinguish toe walking (Le Cras et al., 2011; Alvarez et al., 2007).

A small universal goniometer was used to assess knee and ankle PROM, thigh-foot angle, and transmalleolar angle in both the preliminary and follow-up studies according to the SGL

physical therapy assessment protocol. The universal goniometer has been shown to have high criterion-related validity along with good reliability depending on the joint being assessed; it has been proven more reliable than visual estimation of motion, although intra-tester reliability is stronger than inter-tester reliability (Clarkson, 2005). Watkins, Darrah, and Pain (1995) demonstrated a 4.4° to 6.5° difference between physical therapists in passive ankle DF measurements in children; however, error in intra-tester reliability was reduced by almost half. All passive goniometer measurements were performed in accordance with methods outlined by Clarkson (2005). The primary static clinical measurement of interest for this study was ankle DF PROM; it was assessed in subtalar neutral with the knee flexed to 90 degrees and extended to neutral in a prone, non-weight bearing position. There was no significant correlation between clinical ankle DF and maximum dorsiflexion during stance phase of gait in the preliminary severity classification study; however, this measure was repeated to confirm the finding and to assist in characterizing the participants, particularly with respect to treating community therapists without access to quantitative gait lab data.

3.3.3 Functioning and Disability

Two validated questionnaires were used to assess the activities and participation levels of the ICF as there was not a universal outcome measure that covered the range of ages (14.3-28.8 years) of the participants included in this study. The ICF-CY defines activity as a task or an action carried out by an individual and participation as involvement or experience in life situations (World Health Organization, 2007). In regards to these definitions, the questionnaires primarily measure activity levels with a very small subset of items addressing participation. The two questionnaires both demonstrate a physical and mental component with similar corresponding questions in each domain. Responses from the following questionnaires are all

calculated to normative scores and referenced to the general (healthy) population norm; higher scores denote higher functioning.

Self-reported physical function for adolescents up to 18 years was assessed using the Pediatric Outcomes Data Collection Instrument – a valid and reliable measure of child- and/or parent-reported ability to participate in normal or vigorous daily activities (Klepper, 2011). This questionnaire targets children and adolescents with health problems related to muscle and bone conditions (Appendix E). The PODCI Adolescent Outcomes self-report Questionnaire is an 83-item measure used to assess the following eight scales: upper extremity and physical function, transfer and mobility, sports/physical functioning, pain/comfort, treatment expectations, happiness, satisfaction with symptoms, and global functioning. The Global Functioning Scale is made up of the means of the first four scales and the Happiness Core Scale considers body image items and the ability to keep up with peers. Most items are scored using a four- or five-point Likert scale with one representative of the most positive response (Klepper, 2011). The PODCI has been shown to be reliable, with a Cronbach's alpha range of 0.76-0.95, and a test-retest agreement of 0.71-0.97 (Klepper, 2011).

For young adults over 18 years, self-reported physical function was assessed using the Medical Outcomes Study 36-Item Short Form Survey Instrument – a valid and reliable measure of self-reported functional status in adults with multiple diseases or conditions, including musculoskeletal and neuromuscular conditions (Jenkinson, Wright, & Coulter, 1994). The SF-36 is a 36-item measure used to assess the following eight subscales: physical functioning, role limitations due to physical problems, general health perceptions, vitality, social functioning, role limitations due to emotional problems, general mental health, and health transition (Appendix F). The eight subscales are grouped into a Physical Component Summary and a Mental Component

Summary, corresponding closely to the Global Functioning Scale and Happiness Core Scale in the PODCI, respectively. Items within the subscales are scored using a five-level response weighted Likert scale (Hussey & Harro, 2013). The SF-36 has demonstrated reliability, with a Cronbach's alpha > 0.80 for all dimensions with the exception of the social functioning scale which was 0.76 (Jenkinson et al., 1994).

3.4 Procedures

Data collection included a repeat physical examination and instrumented gait analysis at the SGL at SHHCC in Vancouver and lasted 60 to 90 minutes. Computerized gait data were analyzed retrospectively for each initial gait lab session (1997-2005) and follow-up sessions were collected prospectively (2015-16). All gait analyses consisted of a parent and/or participant interview, lower extremity physical assessment, body mass and height measurement, placement of retro-reflective markers and surface EMG electrodes, and instrumented gait examination. For the initial gait assessments, two video cameras were used to collect walking views from the frontal and sagittal planes on mini digital video cassettes and a third camera collected a close-up view of the feet in the sagittal plane. However, to meet ethics requirements, the motion capture computer was used to collect digital reference video directly to its hard drive for all follow-up assessments. A self-report questionnaire was completed by each participant to assess further any previous or current activity limitations or participation restrictions.

The following motion capture system was used between 1997-2005 to collect baseline data: an 8-camera Motion Analysis system (Motion Analysis Corporation, Santa Rosa, CA), two floor-mounted AMTI force plates (Advanced Mechanical Technology Incorporated, Watertown, MA), modified Helen Hayes marker set (Kadaba, Ramakrishnan, & Wootten, 1990), and

telemetered EMG (Noraxon U.S.A. Incorporated, Scottsdale, AZ) with surface electrode placement over tibialis anterior, medial gastrocnemius, rectus femoris, and semitendinosus/membranosus muscles according to described protocols by Basmajian and Blumenstein (1980). Kinematic data were calculated at 120 Hz using MAC EVA capture software and calculations in OrthoTrak 6.2 Software (Motion Analysis Corporation, Santa Rosa, CA). Kinematic and kinetic data were compared to the OrthoTrak normative database for age.

At follow-up, the master's candidate collected a report of toe walking history and performed all physical therapy assessments including joint ROM, muscle strength, muscle tone, muscle length, lower extremity alignment, lower limb lengths, and gross motor skills, based on current clinical care guidelines (Le Cras et al., 2011). The master's candidate and SGL Biomechanist were blinded to participants' baseline gait data, ITW severity classification, and clinical examination results. Intervention status was confirmed following data processing. The conventional marker set used at baseline and a modified multi-segment foot model defined by Saraswat, MacWilliams, and Davis (2012) were used to guide placement of 63 retro-reflective markers on the participant's skin (Davis et al., 2006; Maurer et al., 2013). Wireless surface EMG electrodes (Delysis Incorporated, Natick, MA) were placed over the same four lower extremity muscles bilaterally, including tibialis anterior, medial gastrocnemius, rectus femoris, and semitendinosus/membranosus. One static standing trial was conducted prior to the walking trials to determine joint centre positions of rotation for the hip, knee, and ankle and to define lower limb segment axes. Three-dimensional position of the markers were tracked with a 12-camera motion capture system (Motion Analysis Corporation, Santa Rosa, CA) using Cortex data collection software while the participant walked at a self-selected typical pace along a ten metre walkway. Ground reaction forces were collected simultaneously from three floor-embedded

force plates located at the middle of the walkway (Advanced Mechanical Technology Incorporated, Allentown, MA). Participants repeated walking trails until 10 clean force plate strikes were collected for each limb; three consistent strides from three different trials were selected for analysis and averaged (Alvarez et al., 2007; Maurer et al., 2013). The representative trials were determined by visual inspection of all traces with the final traces from the session generally the most consistent. Kinematic (120 Hz) and kinetic (1200 Hz) data were calculated using Visual 3D (C-Motion, Germantown, MD) and custom MatLab code.

The change from OrthoTrak to Visual 3D software was designed around the OrthoTrak guidelines for backwards compatibility which have been proven within our gait lab data. Our original single-segment conventional foot model is embedded in the current multi-segment foot model and ankle angle is calculated comparably in each software system. Kinematic and kinetic data were used to determine changes from baseline to follow-up in both non-treatment and treatment groups as well as deviations from normative values. At follow-up, participants were compared to reference values from 34 healthy children assessed in the SGL, aged five to 18 years, comparable to previous OrthoTrak normative values.

3.4.1 Confidentiality

All collected gait and questionnaire data were labeled with non-identifying information to maintain confidentiality and accurate tracking. Data and digital video recordings are stored in the secure, password protected SGL database only accessible by the research team at the SGL. No video recordings were collected from the gait lab video cameras. Participant consent and assent forms as well as completed questionnaires will remain filed in a locked cabinet, in a locked room at SHHCC for a minimum of five years following completion of the study.

3.4.2 Participant Remuneration

There was no reimbursement offered for participation in this study. All adolescents were offered a letter acknowledging their participation in the study to be credited as volunteer hours. Volunteer letters were provided for three participants.

3.5 Data Synthesis and Analysis

Clinical gait data analyses were conducted with parametric, nonparametric and correlation statistics using IBM SPSS Statistics for Windows, Version 23 (IBM Corp., Armonk, NY). All gait data and ankle DF PROM data were checked for normality prior to performing statistical analyses. Knee kinematics, foot orientation, ankle moment data and ankle DF PROM measures were found to be normally distributed. Ankle and pelvis kinematics as well as ankle joint power data and timing of ankle kinematics and kinetics were approximately normal or were found to be mildly skewed. There were no appropriate direct nonparametric tests; therefore, in consultation with a statistician, the decision was made to proceed with parametric testing with an awareness of possible reduction in power to detect change.

To address the first study objective of describing the natural history of ITW and the effect of treatment on long-term outcomes of body functions and structures in adolescents and young adults, a mixed model analysis of variance (ANOVA) was used. Primary dependent variable data analysis compared the trajectory of peak ankle DF during stance at baseline and at follow-up. In secondary data analysis, the mixed model ANOVA was repeated for supporting continuous kinematic variables of ankle angle at initial contact, peak ankle DF in swing, peak knee extension in stance, peak knee flexion in swing, maximum anterior pelvic tilt, maximum foot orientation, ankle kinetics, and ankle DF PROM. The mixed model ANOVA was a 2x2 design

with a third factor; it consisted of one between-subject factor of treatment effect and two within-subject factors of time (two assessment sessions) and side (right and left). The main effects of treatment, time and interactions of treatment over time were determined. The main effect of side was disregarded in the analysis as the differences in sides were simply used to account for twice the data.

The statistical approach of including data from two limbs within a single subject violates the assumption of independence and has been highlighted in orthopaedic research and in studies reporting kinematic and kinetic gait data as a primary outcome measure (Bryant et al., 2006; Park et al., 2010; Saintani, 2010; Sangeux et al., 2013; Niiler & Miller, 2014). Right-left gait data will always be correlated and the implication of reporting on both sides together (pooling foot data) is that groups are considered different more often even when they are not. By overestimating the number of individual data points, the assumption of independence is violated, inflating the type I error rate, thus increasing false positive claims of statistical effects. Alternative approaches for addressing paired data have traditionally included choosing a specific side or a dominant limb; random selection of one side (right or left); or averaging the right-left side data (Sangeux et al., 2013). These methods are overcautious and markedly decrease the degrees of freedom, reducing their power in detecting true differences (Zumbo & Zimmerman, 1991). Another possible statistical approach is Zumbo and Zimmerman's (1991) randomization test for paired data (Zumbo, 1996); it removes the redundancy within pairs, addresses the problem of non-independence, and enables return to the original data to make corrections. For simplicity and the purpose of this project, it was considered most appropriate to place side into the mixed model to enable use of all data points (n=88) without violating the assumption of independent observations.

To address the second study objective of determining the relationship between ITW severity and treatment on long-term outcomes of body functions and structures, a Wilcoxin Signed-Ranks Sum test was used. This nonparametric repeated measure equivalent of a paired difference test was used to accommodate for the ordinal variable of severity classification. The level of ITW severity for each participant was compared from baseline to follow-up and also summarized descriptively.

To address the third study objective of exploring if activity limitations and participation restrictions were demonstrated in these adolescents and young adults, a Pearson's correlation coefficient was conducted. This primary data analysis was used to assess relationships between clinical gait variables and 1) PODCI scores for children up to 18 years or 2) SF-36 scores for young adults over 18 years. Visual analysis of the data was also performed to determine clinical significance. An independent samples t- test was conducted to assess for differences between treatment groups and PODCI or SF-36 scores. The level of ITW severity at follow-up was also compared with PODCI or SF-36 scores using the independent samples t-test.

Finally, to address demographic data, age distribution of participants, time between visits, height, body mass, and body mass index (BMI) were compared between treatment groups with the independent samples t-test. A chi-square test was used to compare sex distribution between treatment groups.

4. RESULTS

4.1 Participant Characteristics

Participant age, time between visits, sex, height, body mass, and BMI are presented in Table 4.1. As described previously, 45 adolescents and young adults were initially recruited; however, the data from one adolescent were excluded from data analysis because of incomplete baseline data. Thus, complete data for 44 participants were analyzed and reported.

Statistically significant differences were found in age at both Gait Lab visits between the non-treatment (NT, concurrent control) and treatment groups. Participants in the NT group were older than those in the treatment group with mean differences of 1.6 years at initial assessment and 2.5 years at follow-up, respectively. Significant differences were also found between groups in height and body mass at the initial visit, and there were significant differences in BMI between groups at both time points.

No significant differences between participant groups were found with respect to length of follow-up between visits and no significant association was found between groups and sex distribution. The average length of time between visits was 13.3 years with a range of 9.4 to 17.8 years over both groups. Sex distribution was approximately equal for the total sample with 21 females and 23 males participating in the study. Although this difference was not significant, it is consistent with the reported incidence of an increased occurrence of ITW in males (Engstrom & Tedroff, 2012; Fox et al., 2006; Eastwood et al., 2000; Stricker & Angulo, 1998), and more males than females received treatment for their toe walking. Table 4.1 outlines participant demographic characteristics by treatment group.

Table 4.1. Participant Demographic Characteristics

	Non-Treatment Group	Treatment Group	p-Value
Age (years)			
Initial visit	8.7 ± 2.7 (4.9-13.0)	7.1 ± 2.2 (4.3-12.2)	0.035*
Follow-up visit	22.6 ± 4.0 (15.8-28.8)	20.1 ± 3.6 (14.3-26.1)	0.032*
Time Between Visits (years)	13.7 ± 2.0 (9.4-16.8)	12.9 ± 2.4 (9.7-17.8)	0.244
Sex (number)			
Total	20	24	0.137
Female	12	9	
Male	8	15	
Height (cm)			
Initial visit	134.3 ± 17.9 (103.0-169.0)	123.1 ± 12.3 (102.9-151.4)	0.018*
Follow-up visit	169.7 ± 9.3 (154.3-186.7)	172.6 ± 11.4 (150.3-199.2)	0.369
Body Mass (kg)			
Initial visit	36.2 ± 15.6 (18.1-68.5)	25.5 ± 7.2 (15.1-44.3)	0.005*
Follow-up visit	78.9 ± 20.2 (45.8-119.7)	69.8 ± 15.9 (53.2-105.6)	0.101
Body Mass Index (kg/m^2)			
Initial visit	19.0 ± 3.6 (14.8-27.3)	16.5 ± 2.3 (13.6-25.0)	0.008*
Follow-up visit	27.3 ± 6.4 (19.2-45.3)	23.4 ± 4.6 (17.2-36.8)	0.025*

Data are given as mean ± standard deviation with minimum-maximum values in parentheses. p-values determined with independent samples t-tests except sex distribution by chi-square test. *Significant difference at $p<0.05$.

A positive family history of ITW was reported in 29% of females and 43% of males over both groups. Fifty seven percent of participants self-reported they still walked on their toes at follow-up; 52% only occasionally and particularly first thing in the morning, in the evening when tired, running, hiking, barefoot, or if they were excited, distracted, anxious, or in a hurry; 5% reported walking on their toes approximately 30-60% of the day and accommodated for this problem with footwear. Participants reported less time spent toe walking following treatment and both groups described toe walking as subsiding or disappearing by the time they entered high school. No adverse effects were reported by participants as a result of treatment.

4.2 Gait Analysis Data

To address the natural history of ITW and the effect of treatment on long-term outcomes of body functions and structures in adolescents and young adults, the results of the mixed model ANOVA were analyzed to determine any significant effects of treatment, time, and interactions of treatment over time. Tables 4.2 and 4.3 outline the results of the statistical analysis of the kinematic and kinetic gait parameters to match those reported most frequently in the ITW literature.

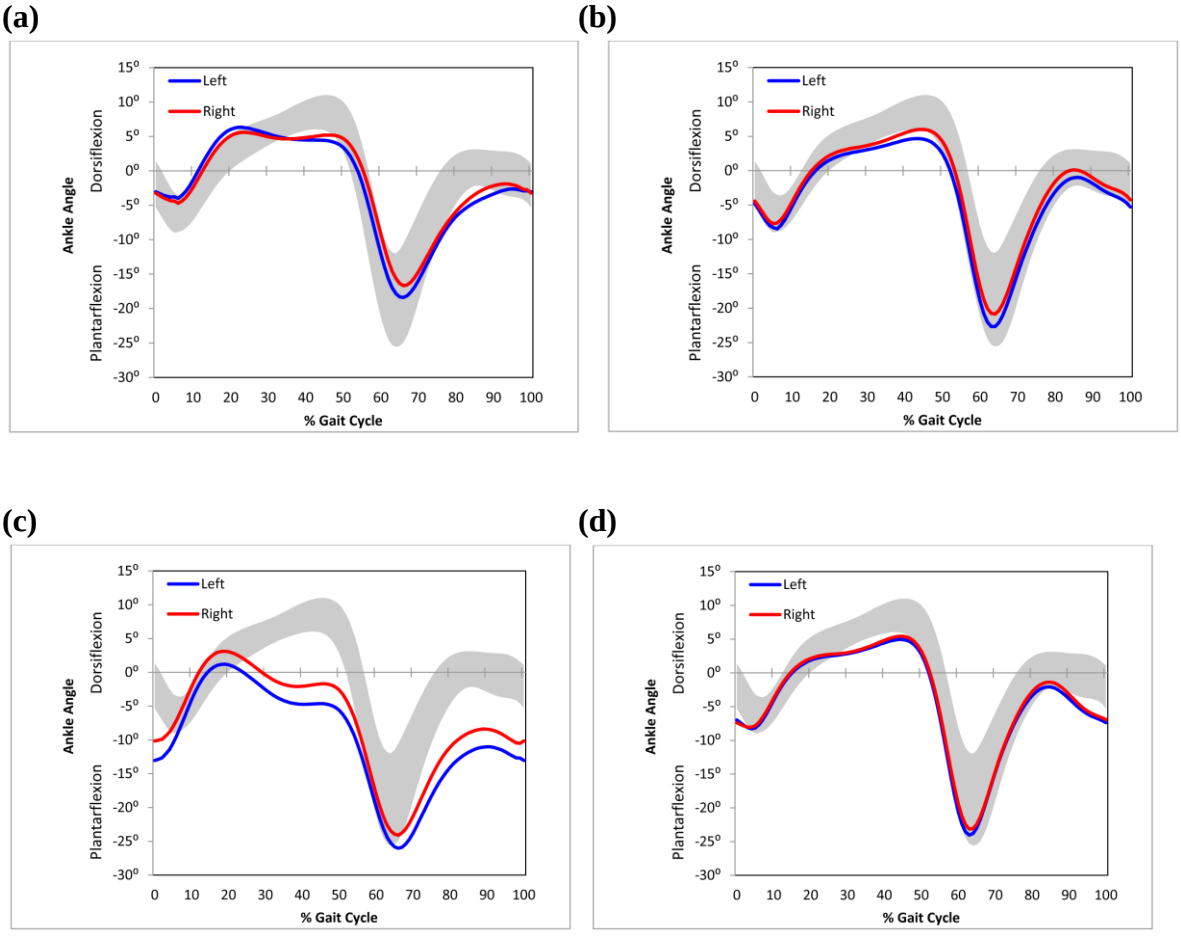
4.2.1 Kinematic Data

Statistically significant differences were found between groups at initial assessment in *all* sagittal plane ankle kinematic variables and their timing in the gait cycle (range of p-values from 0.004 to 0.033). In contrast, at follow-up, no significant differences in ankle kinematic values or timing were observed between groups. Maximum ankle dorsiflexion during swing was the only ankle kinematic variable that did not change significantly over time in either group. Ankle angle at initial contact changed significantly over time in the treatment group only with a mean decrease in plantarflexor angle of 5.8° at follow-up and a small effect size of n^2_p=0.2 (Cohen, 1988).

The primary outcome measure in this study was maximum ankle dorsiflexion angle in stance. Ankle dorsiflexion angle typically increases from approximately 5% to 45% of the gait cycle to a peak value of approximately 10-15° (Perry, 1992; Schwartz, 2004). Peak ankle dorsiflexion in stance in the NT group did not change significantly from baseline to follow-up; however, the timing improved significantly with peak dorsiflexion occurring later and more appropriately in the stance phase of the gait cycle with a small effect size of n^2_p=0.4 (Cohen, 1988). In contrast, ankle dorsiflexion improved over time in the treatment group with a

statistically significant increase of peak dorsiflexion angle by follow-up visit throughout stance with a mean difference and increase of 7.8° and small effect size of n^2_p=0.2 (Cohen, 1988). In addition, in the treatment group, timing of maximum dorsiflexion improved significantly, changing by 10% and occurring later in the gait cycle with a small effect size of n^2_p=0.4 (Cohen, 1988) (Figure 4.1).

Figure 4.1. Graphical Illustration of Sagittal Ankle Kinematic Patterns Grouped by Interventions, (a) NT group at baseline and (b) NT group at follow-up, (c) Treatment group at baseline and (d) Treatment group at follow-up. The gray band represents sagittal ankle kinematics based on normative data within 2 SDs of the mean. The NT group demonstrates a significant change to more appropriate timing of peak ankle dorsiflexion from baseline to follow-up. The treatment group at baseline demonstrates an absent first rocker, abbreviated second rocker, and diminished dorsiflexion during swing. Peak ankle dorsiflexion and timing of this variable improved significantly at follow-up with perseverance of a less severe absent first rocker and mildly abbreviated second rocker.

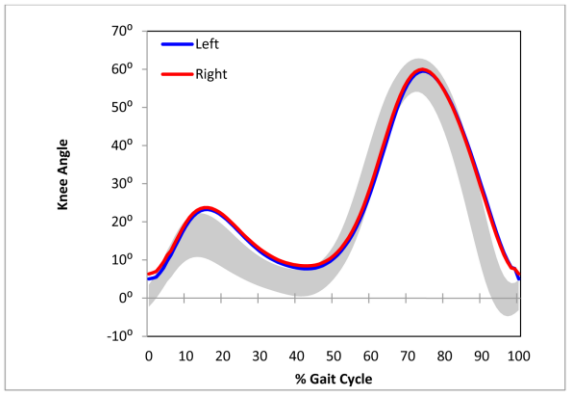


Other reported deviations in gait kinematics in ITW include knee hyperextension, anterior pelvic tilt, and external foot progression. No significant differences between the groups at either initial or follow-up visit were found in knee, pelvis, or foot orientation kinematic variables. Both groups demonstrated similar statistically significant changes over time in peak knee extension in stance, increasing by 7.1° in the NT group and 6.3° in the treatment group, resulting in knee hyperextension at follow-up visit. The knee typically extends to approximately 5° of flexion in mid-stance and does not normally attain end range of motion in gait, much less hyperextension (Schwartz, 2004) (Figure 4.2). Conversely, knee flexion in swing decreased significantly over time in both groups with an 8.4° change in the NT group and a 5.5° change in the treatment group. The effect sizes for this analysis (n^2_p=0.6) exceeds Cohen's (1988) suggestion for a moderate effect.

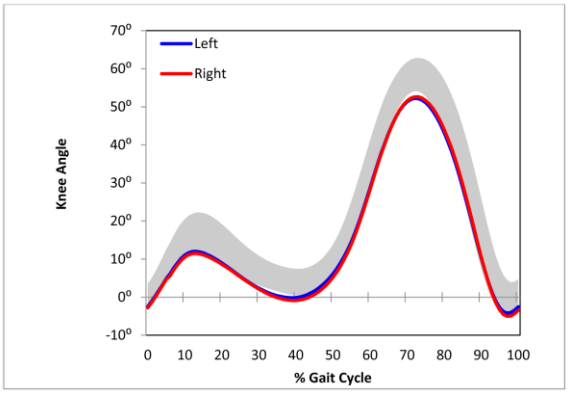
Statistically significantly changes over time were found in maximum pelvic tilt in both groups; participants had a reduction in anterior pelvic tilt between visits; however, the observed changes in degrees were small (effect size of n^2_p=0.1) (Cohen, 1988). No statistically significant effects of treatment, time, or interactions of treatment over time were found in maximum foot orientation, although foot orientation became more external from baseline to follow-up in both groups.

Figure 4.2. Graphical Illustration of Sagittal Knee Kinematic Patterns Grouped by Interventions, (a) NT group at baseline and (b) NT group at follow-up, (c) Treatment group at baseline and (d) Treatment group at follow-up. The gray band represents sagittal knee kinematics based on normative data within 2 SDs of the mean. Peak knee extension in stance is appropriate in both groups at baseline with peak knee extension greater than 2 SDs below SGL normative values at follow-up, indicative of knee hyperextension.

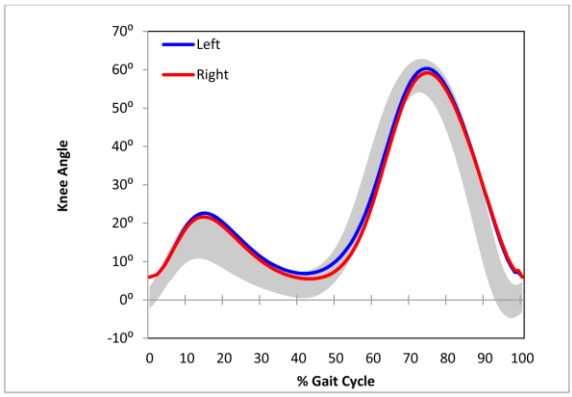
(a)



(b)



(c)



(d)

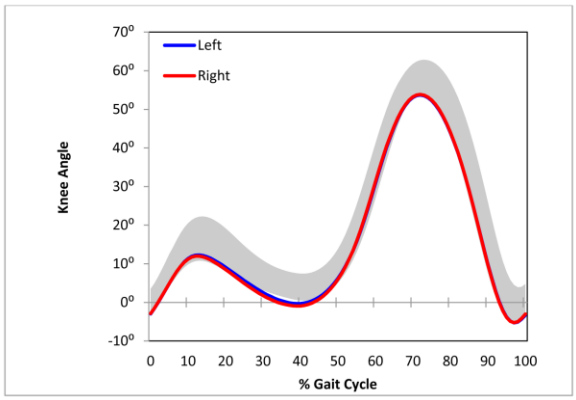


Table 4.2a. Mixed ANOVA Results for Kinematic Parameters in No Treatment Group

Gait Analysis Variable	Initial Visit [95% CI]	Follow-up Visit [95% CI]	p-Value
Ankle angle at initial contact	-3.6 (7.0) [-7.7, 0.4]	-5.0 (3.5) [-7.5, -2.4]	0.449
Peak ankle DF, stance	7.8 (4.2) [4.8, 10.8]	5.8 (4.0) [3.9, 7.8]	0.16
Timing peak ankle DF, first half stance	24.7 (4.3) [22.7, 26.7]	27.9 (4.0) [25.8, 29.9]	0.001*
Timing peak ankle DF, second half stance	40.0 (8.5) [36.8, 43.2]	43.8 (4.7) [41.9, 45.7]	0.046*
Peak ankle DF, swing	-0.68 (4.2) [-2.2, -0.9]	-0.17 (2.4) [-1.6, 1.3]	0.087
Peak knee extension, stance	3.9 (5.0) [1.7, 6.1]	-3.2 (3.2) [-4.9, -1.5]	<0.001*
Peak knee flexion, swing	61.2 (6.2) [58.2, 64.2]	52.8 (3.8) [50.7, 54.9]	<0.001*
Max anterior pelvic tilt	18.9 (5.5) [16.7, 21.2]	17.7 (7.0) [14.6, 20.7]	0.033*
Max foot orientation	-6.9 (8.3) [-10.3, -3.6]	-7.9 (7.9) [-11.0, -4.8]	0.193

Angle variables are in degrees with values given as mean (standard deviation).
Ankle dorsiflexion positive values, plantarflexion and external foot orientation negative values.
Timing variables are described in % of gait cycle.
*Significant difference at $p<0.05$.

Table 4.2b. Mixed ANOVA Results for Kinematic Parameters in Treatment Group

Gait Analysis Variable	Initial Visit [95% CI]	Follow-up Visit [95% CI]	p-Value
Ankle angle at initial contact	-13.2 (10.7) [-16.9, -9.5]	-7.4 (7.2) [-9.8, -5.1]	0.001*
Peak ankle DF, stance	2.4 (8.4) [-0.3, 5.2]	6.2 (4.9) [4.4, 7.9]	0.006*
Timing peak ankle DF, first half stance	20.9 (4.8) [19.1, 22.7]	26.9 (5.1) [25.0, 28.7]	0.001*
Timing peak ankle DF, second half stance	34.1 (7.5) [31.2, 37.0]	44.1 (4.7) [42.3, 45.8]	<0.001*
Peak ankle DF, swing	-3.6 (3.6) [-5.0, -2.2]	-1.6 (4.1) [-2.9, -0.3]	0.087
Peak knee extension, stance	2.8 (5.5) [0.8, 4.8]	-3.5 (4.5) [-5.0, -1.9]	<0.001*
Peak knee flexion, swing	60.3 (7.6) [57.6, 63.0]	54.8 (5.4) [52.9, 56.7]	<0.001*
Max anterior pelvic tilt	17.6 (4.6) [15.6, 19.7]	14.8 (6.6) [12.0, 17.6]	0.033*
Max foot orientation	-5.8 (7.6) [-8.9, -2.7]	-7.4 (6.8) [-10.2, -4.6]	0.193

Angle variables are in degrees with values given as mean (standard deviation).
Ankle dorsiflexion positive values, plantarflexion and external foot orientation negative values.
Timing variables are described in % of gait cycle.
*Significant difference at $p<0.05$.

4.2.2 Kinetic Data

The kinematic parameters analyzed thus far assist in describing the motion occurring at the ankle (as well as the knee and pelvis) over the gait cycle. Further analysis of sagittal plane ankle kinetics augments understanding of this motion by measuring power produced by the muscles around the ankle. The key kinetic parameters analyzed in this study were peak internal ankle moment in stance and peak ankle power generation in the final 20% of stance. Peak ankle moment in stance is further divided into maximum values in the first and second half of stance and timing as a percentage of the gait cycle for additional interpretation of the early ankle moment or double bump pattern seen frequently in ITW.

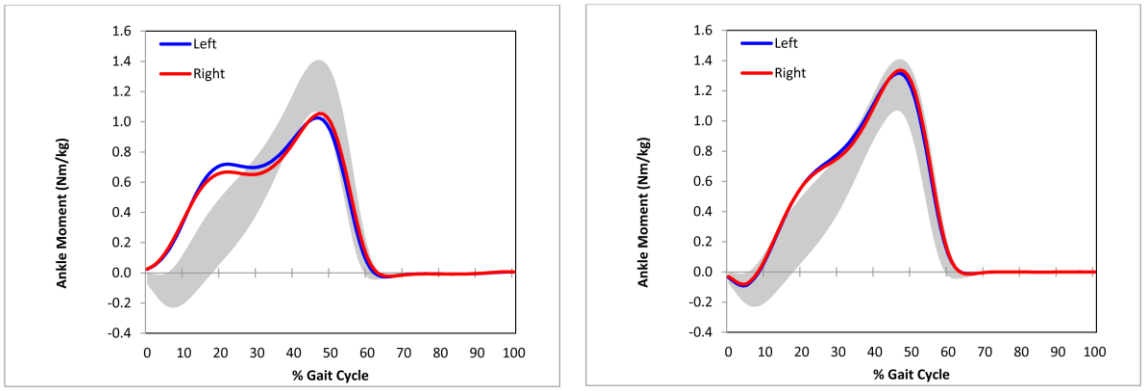
Overall, a statistically significant difference in peak internal ankle plantarflexor moment was found in stance in both groups over time. Participants in both groups changed in a similar way with the peak plantarflexor moment increasing between initial visit and follow-up with a medium effect size of n^2_p=0.5 (Cohen, 1988). The peak ankle plantarflexor moment occurs typically at approximately 50% of the gait cycle (Schwartz, 2004), during the second half of stance. In this latter half of the stance phase; however, the NT and treatment groups change over time in different ways. At the initial visit, the treatment group had a lower maximum ankle plantarflexor moment than the NT group, yet attained a higher maximum ankle plantarflexor moment at follow-up than the NT group, with a large effect size of n^2_p=0.8 (Cohen, 1988). Timing of peak ankle plantarflexor moment also changed significantly in the treatment group by occurring later and more appropriately in the second half of stance with a small effect size of n^2_p=0.1 (Cohen, 1988).

Statistically significant differences were found between the groups at initial assessment in peak ankle plantarflexor moment in the first half of stance (p=0.004) and in the timing in the gait cycle (p=0.001) with no significant differences between groups at follow-up in either parameter. The peak ankle plantarflexor moment in the first half of stance remained the same between visits in the NT group, yet timing improved significantly and occurred later in the gait cycle with a medium effect size of n^2_p=0.6 (Cohen, 1988). Conversely, treatment group participants demonstrated a statistically significant decrease in peak ankle plantarflexor moment in the first half of stance between visits with more appropriate timing of 8.5% later in the gait cycle with small (n^2_p=0.1) and medium (n^2_p=0.6) effect sizes, respectively (Cohen, 1988). However; peak ankle moments in the first half of stance remained atypical, greater than two standard deviations

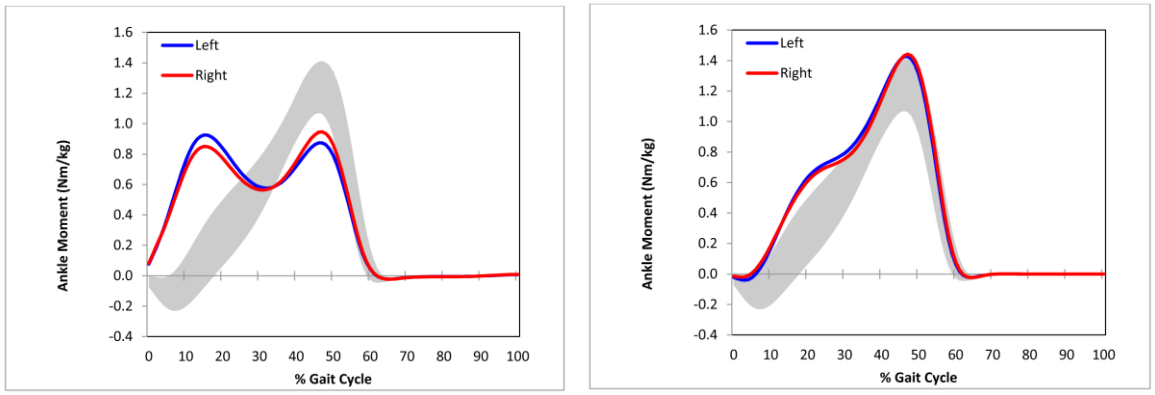
above the SGL normative values, in 95% of the NT group and 91% of the treatment group with 89% of all participants reflecting perseverance in early heel rise to a varying extent (Figure 4.3).

Figure 4.3. Graphical Illustration of Sagittal Ankle Kinetic Patterns Grouped by Interventions, (a) NT group at baseline and (b) NT group at follow-up, (c) Treatment group at baseline and (d) Treatment group at follow-up. The gray band represents sagittal ankle kinetics based on normative data within 2 SDs of the mean.

(a) **(b)**



(c) **(d)**



The internal peak ankle plantar flexor moment combined with increased angular velocity at the ankle produces a burst of power generation at the ankle starting at about 40% of the gait cycle and increasing to its maximum value at approximately 50% of the gait cycle (Schwartz, 2004). Like the results of peak internal ankle plantarflexion moment in the second half of stance, peak ankle power generation in the final 20% of stance changed over time in different ways between the groups. No significant differences were found between groups at initial assessment;

yet, they changed and were significantly different by their follow-up visit. At the initial visit, the treatment group had less peak ankle power generation than the NT group, but attained higher maximum ankle power generation at follow-up than the NT group, with an effect size (n^2_p=0.7) exceeding Cohen's (1988) suggestion for a moderate effect.

Table 4.3a. Mixed ANOVA Results for Kinetic Parameters in No Treatment Group

Gait Analysis Variable	**Initial Visit [95% CI]**	**Follow-up Visit [955 CI]**	**p-Value**
Peak ankle PF moment, stance	1.1 (0.2) 1.2 [1.0, 1.2]	1.4 (0.1) [1.3, 1.4]	<0.001*
Peak ankle PF moment, first half stance	0.8 (0.3) [0.7, 0.9]	0.8 (0.2) [0.7, 0.8]	0.871
Timing peak ankle PF moment, first half stance	24.7 (5.2) [22.3, 27.1]	28.8 (3.4) [26.9, 30.7]	0.001*
Peak ankle PF moment, second half stance	1.1 (0.2) [1.0, 1.1]	1.3 (0.1) [1.3, 1.4]	<0.001*
Timing peak ankle PF moment, second half stance	47.1 (2.9) [45.3, 48.8]	47.8 (1.4) [47.3, 48.3]	0.350
Peak ankle power generation, final 20% stance	1.7 (0.4) [1.5, 1.9]	2.4 (0.4) [2.2, 2.6]	<0.001*

All moment variables are in Nm/kg and are from an internal frame of reference.
Timing variables are described in % of gait cycle.
Ankle power generation is in W/kg.
*Significant difference at p<0.05.

Table 4.3b. Mixed ANOVA Results for Kinetic Parameters in Treatment Group

Gait Analysis Variable	Initial Visit [95% CI]	Follow-up Visit [95% CI]	p-Value
Peak ankle PF moment, stance	1.2 (0.3) [1.1, 1.2]	1.3 (0.2) [1.4, 1.5]	<0.001*
Peak ankle PF moment, first half stance	1.1 (0.3) [0.9, 1.2]	0.8 (0.2) [0.8, 0.9]	0.001*
Timing peak ankle PF moment, first half stance	18.8 (6.0) [16.6, 21.0]	27.3 (4.9) [25.6, 29.0]	<0.001*
Peak ankle PF moment, second half stance	0.9 (0.2) [0.9, 1.0]	1.4 (0.2) [1.4, 1.5]	<0.001*
Timing peak ankle PF moment, second half stance	45.9 (6.4) [44.3, 47.5]	48.1 (1.3) [47.6, 48.6]	0.005*
Peak ankle power generation, final 20% stance	1.4 (0.6) [1.3, 1.7]	2.7 (0.7) [2.5, 2.9]	<0.001*

All moment variables are in Nm/kg and are from an internal frame of reference.
Timing variables are described in % of gait cycle.
Ankle power generation is in W/kg.
*Significant difference at $p<0.05$.

4.2.3 Electromyographic Data

Electromyographic data were considered in brief to support the corresponding ankle kinematic and kinetic results. There was preparatory firing of gastrocnemius in swing in 20% of participants in the NT group and 45.8% of participants in the treatment group at initial assessment. By follow-up visit, there was no preparatory firing in NT participants and preparatory firing in only 12.5% of participants in the treatment group.

4.2.4 ITW Severity Classification

To determine the relationships between ITW severity and treatment on long-term outcomes of body functions and structures in adolescents and young adults, the level of ITW severity for each participant was compared from initial visit to follow-up and summarized descriptively. The results are illustrated in Table 4.4. A Wilcoxin Signed-Ranks Sum test was also used to examine group differences and found a statistically significant difference in severity

from baseline to follow-up in the treatment group ($p<0.001$) compared to a non-significant difference in the NT group ($p=0.083$).

In the NT group, 67% of six participants classified as mild toe walkers at their initial visit remained at the same level of severity at follow-up. Thirty three percent of these participants changed to type 2, or moderate, toe walkers at follow-up. Of the 12 participants classified as moderate toe walkers at initial visit, 42% improved to become mild toe walkers at follow-up and 58% remained at the same level of severity. Only two participants were classified as severe toe walkers in the initial visit and one improved to mild and the other to moderate toe walking by follow-up. Overall in the NT group, 35% of participants improved, 55% of participants remained the same and 10% of participants declined in level of severity over time.

In the treatment group, no participants were classified as mild toe walkers at initial visit, and of the nine participants classified as moderate toe walkers, 33% improved to become mild toe walkers at follow-up and 67% remained at the same level of severity. As expected, the majority of participants who received treatment were classified as severe toe walkers at initial visit – 33% changed to mild toe walkers and 67% changed to moderate toe walkers at follow-up. In the treatment group, 75% of participants improved and 25% remained the same with no participants classified as severe toe walkers at follow-up.

Table 4.4. Idiopathic Toe Walking Severity Classification

	No Treatment (Control)		**Treatment**	
Severity Group	**Initial Visit**	**Follow-up Visit**	**Initial Visit**	**Follow-up Visit**
Type 1 Mild	6	10	0	8
Type 2 Moderate	12	10	9	16
Type 3 Severe	2	0	15	0

All values are reported as number of participants.

4.3 Clinical Range of Motion

Clinical passive ankle dorsiflexion range of motion was analyzed with a mixed model ANOVA to determine any significant effects of treatment, time, and interactions of treatment over time (Table 4.5).

A similar statistically significant decrease in ankle dorsiflexion PROM, measured with the knee flexed to 90°, was found in both groups from initial to follow-up visit with a medium effect size of n^2_p=0.5 (Cohen, 1988). Ankle dorsiflexion PROM measured with knee extension also decreased over time; however, while the treatment group had less ankle dorsiflexion at initial visit, they maintained more dorsiflexion over time than the NT group. The effect size was small (n^2_p=0.2) (Cohen, 1988). No significant difference between groups was found at either initial visit or follow-up for both measurements of PROM. Although maximum ankle dorsiflexion in stance in the NT group also decreased over time between visits, the change was not significant (7.8°±4.2 – 5.8°±4.0) and there were increased dorsiflexion angles in stance compared to clinical assessment. Conversely, the treatment group demonstrated a statistically significant increase in maximum ankle dorsiflexion in stance over time (2.4°±8.4 – 6.2°±4.9) compared to the decrease in passive ankle dorsiflexion measured clinically.

Table 4.5. Clinical Passive Ankle Range of Movement

	No Treatment (Control)			Treatment		
Variable	**Initial Visit**	**Follow-up Visit**	**p-Value**	**Initial Visit**	**Follow-up Visit**	**p-Value**
Ankle dorsiflexion, knee extended	3.9 (6.2) [0.8, 6.9]	-0.6 (2.9) [-2.1, 0.8]	0.001*	2.5 (7.9) [-0.3, 5.3]	0.2 (3.8) [-1.1, 1.6]	0.001*
Ankle dorsiflexion, knee flexed	7.4 (5.7) [4.8, 9.9]	1.3 (2.8) [-0.4, 2.9]	<0.001*	5.6 (6.1) [3.3, 8.0]	0.8 (4.3) [-0.7, 2.3]	<0.001*

Angle variables are in degrees with values given as mean (standard deviation).
*Significant difference at p<0.05; 95% Confidence Intervals shown in [ ].

4.4 Functioning and Disability Data

Finally, to explore for potential activity limitations and participation restrictions in this sample, self-reported physical function was assessed using the PODCI and SF-36 questionnaires, for adolescents up to 18 years and young adults over 18 years, respectively, and summarized descriptively. The scores were analyzed with Pearson's correlation coefficient to explore associations between gait parameters and activity limitation or participation restrictions. No positive correlations were found between any gait parameters or ankle DF PROM and questionnaire scores. Independent samples t-tests were also used to assess for differences between treatment and severity groups and questionnaire scores. No significant differences were found between treatment or severity and either scale or component of the two questionnaires.

The PODCI was completed by 13 adolescents (38% female, 62% male) between the ages of 15.0 to 18.8 years. In the Global Functioning Scale, 54% of participants of the combined groups scored above the general population mean, 8% scored at the general population mean and 38% scored below. In the Happiness Scale, 54% of participants scored above the general population mean and 46% scored below the general population mean. No statistically significant correlations were found between gait variables or passive ankle dorsiflexion range of motion at follow-up visit and questionnaire scores.

The SF-36 was completed by 31 young adults across both groups (52% female, 48% male) between the ages of 19.8 to 28.0 years. In the Physical Component Summary, 68% of participants scored above the general population mean, 29% scored at the general population mean and only 3% scored below. In the Mental Component Summary, 29% of participants scored above the general population mean, 42% scored at the general population mean and 29%

scored below. No statistically significant correlations were found between gait variables or passive ankle dorsiflexion range of motion at follow-up visit and questionnaire scores.

Further, 61% of all participants self-reported no activity or participation limitations. The remaining participants reported the following limitations and/or symptoms: repetitive ankle sprains (11%); lower extremity muscle tightness (9%); difficulty with stairs, hills, hiking and/or running (9%); foot pain (7%); difficulty with balance and/or squatting (7%); carrying something heavy upstairs (2%); difficulty downhill skiing (2%); and diminished endurance for walking long distances (2%).

5. DISCUSSION

5.1 Summary and Discussion of Results

This descriptive, explorative study assessed a cohort of adolescents and young adults with a history of idiopathic toe walking to determine the natural course of toe walking; implications of treatment and severity on long-term outcomes of body functions and structures; and associated activity limitations or participation restrictions. Natural history studies and long-term outcomes are limited in the ITW literature with few considering the impact of intervention on more than one domain of the ICF-CY framework. Forty-four participants were grouped by treatment or absence of structured treatment and underwent repeat three-dimensional clinical gait analysis at a mean of 11.8 years post-intervention or mean of 13.7 years post-baseline gait assessment, respectively.

Twenty children receiving no reported treatment for ITW, other than recommendations for incorporating stretching into their activities of daily living, demonstrated significant improvements in timing of ankle kinematics, improved ankle moments, and power as adolescents and young adults at long-term follow-up. Sagittal ankle kinematics and severity did not change significantly over time in this group of predominantly mild and moderate toe walkers. These results were considered to reflect the natural history of toe walkers included in this study. Twenty-four participants treated, primarily conservatively, for ITW as children showed significant improvements in ankle kinematics, ankle kinetics, and ITW severity at long-term follow-up. Irrespective of the increase in peak ankle DF seen during gait in the treatment group, clinical passive ankle DF became more restricted over time in both groups. Compensatory knee hyperextension was observed in both groups at follow-up with atypical internal ankle moment patterns remaining in 89% of all participants, suggestive of perseverance of some degree of toe

walking. Participant questionnaire scores reflecting activity and participation levels across groups were above the general population mean for global physical function.

The natural history of ITW has been described in only two other studies in the literature (Eastwood et al., 2000; Stricker & Angulo, 1998). Stricker and Angulo (1998) demonstrated that 48 children aged two to 13 years at initial assessment with mild heel-cord contractures (10° median passive ankle DF) and untreated for ITW showed no change in passive ankle DF at mean follow-up of 3 years (range, two to eight years) with only 25% parent satisfaction regarding their child's gait. No gait data was obtained, but parent satisfaction levels indicated that toe walking persisted in 75% of the children not receiving intervention. In a similar study, Eastwood et al. (2000) observed 49 children untreated for toe walking and aged 1.5 to 10 years at presentation with a mean follow-up of 3 years (range, two to 12 years). They found that although there was a statistically significant improvement in time spent walking on toes, toe walking persisted in 88% of children as per physician-determined outcomes undertaken using wet footprint analyses. Signs of toe walking also persisted in the majority of participants in the current study representing untreated ITW. In contrast, the sample size in the current study was smaller; length of follow-up time was longer; participant age was higher at initial assessment and mean passive ankle DF was lower (mean 3.9°) at baseline and did not remain static but declined over time to -0.6°.

The two retrospective cohort studies addressing the natural history of ITW also compared untreated ITW with results following below-knee casting or solid AFO treatment in 17 children (Stricker & Angulo, 1998) and serial below-knee casting in 41 children (Eastwood et al., 2000). The premise of casting with the muscle in a lengthened position is based on the addition of sarcomeres to the muscle fibres (i.e., of the gastrocnemius and/or soleus muscle/s) and the concurrent stretch of the noncontractile elements (i.e., Achilles tendon) (Brouwer et al., 2000;

Gossman, Sahrmann, & Rose, 1982). These studies found no long-term significant differences in toe walking (Eastwood et al., 2000) or median passive ankle DF and parent satisfaction regarding gait (Stricker & Angulo, 1998). The mean follow-up for the serial casting group was 3.7 years (range, two to 21.5 years), differing slightly from the duration of follow-up for the untreated group in this study (Stricker & Angulo, 1998). These results are in contrast to the beneficial and sustained results found in gait kinematics, kinetics, and severity in the current study, although a lack of gait normalization is consistent across studies and passive ankle DF declined significantly in our study.

The use of BoNT-A in the treatment of ITW has also been described in combination with either physical therapy treatment (Brunt et al., 2004; van Bemmel et al., 2012; Sätilä et al., 2016) or casting (Engström et al., 2013; Jacks et al., 2004). Brunt et al. (2004) showed that timing of gastrocnemius activity and duration of tibialis anterior activity were more appropriate following BoNT-A injections and were associated with improving initial contact toward flatfoot or heel contact patterns up to one year. Engström et al. (2010), in their original BoNT-A study, demonstrated similar results with improved walking pattern, but lack of complete cessation of toe walking, also up to one year. In contrast, a recent RCT found that the addition of BoNT-A injections to conservative treatment did not significantly improve toe walking at two year follow-up (Sätilä et al., 2016). Both Brunt et al. (2004) and Engström et al. (2010) suggested that the addition of serial casting following BoNT-A injections may be necessary to improve gait outcomes. However, further research by Engström et al. (2013) demonstrated that the addition of BoNT-A prior to casting did not significantly improve gait outcomes of cast-only treatment at one year. Gait patterns improved in both treatment groups reflecting improved ankle angle at initial contact, an increase in peak ankle DF during stance of approximately 9° with improved

timing in the gait cycle, and greater peak ankle power generation; overall, severity improved significantly by follow-up with utilization of the same severity classification (Alvarez et al., 2007). The results of the present study parallel these treatment outcomes; albeit with outcomes demonstrating a more modest sustained improvement in peak ankle DF during stance of almost 4°, possibly reflective of the longer term follow-up duration. In contrast, where Engström et al. (2013) found less knee hyperextension in midstance and increased knee flexion in swing one year following treatment, the present study demonstrated greater knee hyperextension and diminished knee flexion in swing at mean follow-up of approximately 11 years. Compensatory knee hyperextension may increase with time, although the diminished knee flexion in swing is not congruent with McMulkin et al.'s (2016) postulation that increased peak knee flexion during swing is necessary for foot clearance once individuals are no longer forefoot weight bearing.

Systematic review of the ITW literature suggests there is not an obvious long-term benefit of serial casting or surgical treatment for children with persistent toe walking apart from improved passive ankle DF for those treated surgically (van Bemmel et al., 2014; van Kuijk et al., 2014). Precursory evidence based on a limited number of RCTs and a large number of retrospective cohort studies indicate short-term improvements in passive ankle DF, gait parameters, and toe walking for both interventions. Few studies report on long-term outcomes. Stricker and Angulo (1998) found that 15 children who underwent triceps surae lengthening maintained significantly improved ankle DF PROM at mean 3 year follow-up with 67% parental satisfaction of child's gait, indicative of persistent toe walking in 33% of participants. Eastwood et al. (2000) showed that 46 children treated surgically had significantly reduced time spent toe walking with persistent toe walking in 63% at mean 7.9 year follow-up. Stott et al. (2004) reported the results of 13 adolescents and young adults treated as children for ITW with serial

casting (*n*=6) and surgery (*n*=7) who underwent gait analysis a mean of 10.8 years post-intervention. Persistent changes were seen in ankle kinematics and kinetics with second rocker formation atypical in 92% of participants. Similarly, second rocker formation remained atypical in both treatment groups at follow-up in the current study. Stott et al. (2004) found mean peak ankle DF during stance was 9° with timing occurring at less than 50% of the gait cycle (and less than 25% in two participants).

In their recent study evaluating gait outcomes greater than five years following surgery, McMulkin et al. (2016) found similar results in mean peak ankle DF during stance (8.9°). Significant improvements were seen in mean pelvic tilt, mean peak ankle DF in stance and swing, the Gait Deviation Index, ankle moment, and ankle power five years post-operatively. Participants showed increased tightness of ankle DF PROM with knee extension between one and five year follow-up. Both studies found no relationship between passive ankle DF in physical exam and peak ankle DF during gait which was also confirmed in the present study. In the present study, the mean peak ankle dorsiflexion in stance at follow-up was less than the previous studies reported, at 6.2°. Passive ankle DF with the knee extended diminished from 2.5° at baseline to 0.2° at 11.8 years mean follow-up in the present study compared to the change from -8.7° pre-operatively to 0.6° five years post-operatively found by McMulkin et al. (2016). Stott et al. (2004) did not have passive ankle DF data pre-treatment, but at 10.8 years mean follow-up, there were 9° and 6° in the cast and surgical groups, respectively. The reduced passive ankle DF could partly be attributed to the documented decrease in ankle joint complex ROM that occurs after 14 to 17 years of age (Grimston et al., 1993).

Finally, this study is the first to report that after standardized evaluation, adolescents and young adults with a history of ITW demonstrate activity levels and a small subset of participation items within normal limits. Of note is that McMulkin et al. (2016) comment descriptively that all eight subjects in their study were unrestricted in activities.

5.2 Limitations

This study has several limitations. Complete baseline data was not available with one potential participant and precluded the use of their follow-up data. Data collection was limited by the number of participants willing to return for long-term follow-up and a lack of control over the treatment received since their initial assessment. As a result, comparison was only possible between reported untreated ITW and toe walking treated by serial casting with or without preceding BoNT-A injections. Ideally, comparison would have been possible with a group of adolescents and young adults treated surgically for ITW as children. True comparison of the natural history of ITW and outcomes following casting was not possible as the two groups had a number of significant differences at baseline including age, body mass, and BMI. Ankle kinematic data was also significantly different at baseline in the majority of parameters, limiting further comparison.

Another limitation of this study is the use of the non-validated classification scale developed previously to determine severity in ITW (Alvarez et al., 2007). Nevertheless, recent use of the scale by several other authors enabled limited comparison among similar studies (Engstrom et al., 2010, 2013; McMulkin et al., 2016; Herrin & Geil, 2016). The recognition of a spectrum in the severity of ITW was useful in considering outcomes of both untreated and treated toe walking. Other authors have critiqued the classification system as limited because of

its requirements for motion analysis equipment which are not common in many clinics or outpatient departments (Le Cras at al., 2011). Also, it was necessary to use two validated questionnaires to assess the activities and participation levels of the ICF-CY as there was not a universal outcome measure that covered the range of ages of the participants included in this study. However, the two questionnaires both demonstrate physical and mental health components with similar corresponding questions in each domain. Responses from both questionnaires are calculated to normative scores and referenced to general (healthy) population means. Both the PODCI and the SF-36 are limited in terms of their ability to measure participation, particularly the SF-36, thus drawing conclusions regarding the effects of treatment on participation is limited.

Statistically, ankle and pelvis kinematics, ankle joint power data, and timing of gait parameters were only approximately normal or found to be mildly skewed in some cases. This potentially reduces the power to detect change in the variables being evaluated. ANOVA; however, is considered to be reasonably robust to deviations from normality due in part to the central limit theorem, which suggests that as sample size increases, the subsequent sample mean will come closer to representing a normal distribution (Gamst, Meyers, & Guarino, 2008).

5.3 Implications for Future Research

The identification of conservative treatment methods to optimize gait and limit secondary compensations in children, adolescents and young adults with ITW is an important area for future research. As young adults grow older, it will be important to continue to follow their outcomes to determine truly long-term sequelae of ITW. To further investigate ideal treatment strategies, larger samples, high-quality of evidence studies, and RCTs are necessary to increase

the understanding of factors influencing treatment outcomes for idiopathic toe walkers. Larger samples will allow for more robust statistical results and appropriate power to determine real change. Results from present, recent high-quality studies and evidence-based guidelines should be integrated into practice and monitored consistently for outcomes. In future studies, all levels of severity and treatment should be equally represented to better understand prognosis and necessity of treatment for ITW.

5.4 Implications for Practice

This study is the first to use three-dimensional computerized gait analysis to determine the natural history of idiopathic toe walking and the long-term treatment outcomes following casting +/- BoNT-A. It provides outcomes based on gait parameters, severity, range of motion data, and self-report questionnaires. The results of the current study highlight potential conservative measures to avoid unnecessary surgical intervention. Given that children who have undergone surgery for ITW continue to demonstrate abnormal gait patterns, physical therapists, orthotists, and orthopedic surgeons need to be able to provide improved, evidence-based education to children and their families in terms of operative decision-making. The results of this study and the higher-level evidence studies discussed in the systematic review will potentially reduce or prevent inherent costs in terms of discomfort, pain, time and specific expenses for children and their families.

5.5 Conclusion

The natural history of toe walking suggests that children with mild or moderate ITW severity left untreated may still gain significant improvements in timing of ankle kinematics, improved ankle moments, and power by the time they reach adolescence and/or young

adulthood. Children with moderate and severe ITW treated primarily conservatively with casting may show significant improvements in ankle kinematics, ankle kinetics, and severity as adolescents and/or young adults. Clinical passive ankle DF will likely become more restricted over time in both groups, irrespective of an expected increase in peak ankle DF during stance in those receiving treatment. Compensatory knee hyperextension and atypical ankle moment patterns are likely in both groups at follow-up, suggesting perseverance of toe walking at some level. Despite these anatomical, kinematic, and kinetic limitations, adolescents and young adults with a history of ITW show minimal limitations in activity levels and minimal participation restrictions across groups.

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APPENDICES

Appendix A: Participant Information Letter

Information Letter
Long-term Follow-up of Idiopathic Toe Walking

May 8, 2015

To << Participant and Parent Names>>,

I am sending you this information package to tell you about a new research project that is being done at the Shriners Gait Lab at Sunny Hill Health Centre for Children. It might be something that you are interested in learning more about. [redacted] and her research team are conducting this study and you or your child is being invited to participate because you or your child was once diagnosed with idiopathic toe walking. By conducting this study, we hope to increase our knowledge regarding the outcome of this diagnosis. You or your child is invited to attend a repeat Gait Lab assessment at Sunny Hill because you or your child was once assessed and/or treated for toe walking and we would like to study how you walk 10 years (or more) following this last visit.

Some children walk on their toes when they are really little, and others keep walking on their toes when they grow older. We are doing this study to find out how many children that were seen at the Gait Lab are still toe walking now that they are older children or young adults.

In the event that you or your child is no longer residing at the address as listed at the Shriners Gait Lab at Sunny Hill Health Centre for Children, the current resident will be asked to provide a forwarding address, if they have one. This information letter will then be mailed to the appropriate address.

Please read the attached information and consent/assent form(s).

You will also receive a phone call in about 2 weeks from the project's co-investigator, [redacted]. She will tell you more about the study and answer your questions. You can decide then if you want to be part of this study.

If you decide now that you **do not** want to hear more about this study that is fine as well. It will not affect your care at Sunny Hill Health Centre for Children in any way. To tell us that you do not want to hear more about the study, you can do the following:

You can phone and leave a message with your name and your child's name and the name of this study (Idiopathic Toe Walking) for [redacted], at Sunny Hill Health Centre for Children ([redacted]) or by email to [redacted]. Just say that you do not want to be contacted about the Idiopathic Toe Walking study. **If you do this, you will not receive any more information about the study.**

Your participation is entirely voluntary, and it is up to you to decide whether or not to take part in this study. If you do not wish to participate, you do not have to provide any reason for your decision, nor will you lose the benefit of any medical care now or in the future. If you or your child has any questions or desire further information with respect to this study, please contact the principal investigator, [redacted], at [redacted].

Thank you for taking the time to review the attached information.

Sincerely,

[redacted]

Appendix B: Consent Form

PARTICIPANT INFORMATION AND CONSENT FORM
Long-term Follow-up of Idiopathic Toe Walking

Principal Investigator:

Co-Investigators:

Sponsors: Funding for this project is provided by graduate awards from the Canadian Institutes of Health Research (CIHR) and the University of British Columbia.

If you are a parent or legal guardian of a child who may take part in this study, permission from you and the assent (agreement) of your child may be required. When we say "you" or "your" in this consent form, we mean you and/or your child; "we" means the doctors and other staff.

1. Invitation to participate

You are invited to participate in this study because you were diagnosed and/or assessed for toe walking from 1997-2005. Children and young adults with a history of toe walking will participate in this study at Sunny Hill Health Centre for Children. The goal of this study is to determine the natural history of toe walking at least 10 years following orthopaedic assessment and gait analysis. In addition, we hope to gather information that will help us to develop appropriate guidelines for treating children who toe walk.

2. Your participation is voluntary

Your participation is voluntary. You have the right to refuse to participate in this study. If you decide to participate, you may still choose to withdraw from the study at any time without any negative consequences to the medical care, education, or other services to which you are entitled or are presently receiving.

You should be aware that there is a difference for both you and your doctor between being a patient and being a research participant. As a patient all medical procedures and treatments are carried out for your benefit only according to standard accepted practice. As a research participant you and your doctor also must take into account the requirements for the research study. These may include procedures and treatments that are not part of standard practice or are not yet proven. This consent form describes the diagnostic and treatment procedures that are being carried out for research purposes. Please review the consent document carefully when deciding whether or not you wish to be part of the research and sign this consent only if you accept being a research participant.

If you wish to participate in this study, you will be asked to sign this form.

Please take time to read the following information carefully and to discuss it with your family, friends, and doctor before you decide.

3. Who is conducting this study?

This study is being conducted by [redacted]. [redacted] will be conducting the study as a requirement for the completion of her Master of Science thesis for the University of British Columbia. [redacted] has been sponsored by a CIHR Canada Graduate Scholarship – Master's Award and the University of British Columbia for her graduate studies. Findings from this study will be submitted for publication in a peer-reviewed journal. None of the investigators of this study will receive payment for enrolment of participants or from the results of this study.

4. Background

When children learn to walk, they often walk on their tiptoes. Those who walk on their toes for several years with no underlying medical reason have a condition called idiopathic toe walking

(ITW). Idiopathic means that we do not know why something occurs. ITW may be associated with pain in the legs or feet, frequent tripping or falling, and ankle injuries. Some children naturally outgrow this walking pattern; some develop an adapted foot position, whereas other children develop shortened calf muscles.

Children with ITW are typically assessed in the Orthopaedic clinic at BC Children's Hospital and if the toe walking persists, they are referred to the Shriners Gait Lab at Sunny Hill Health Centre for Children to further assess how they walk. In the Gait Lab, foot pressures and movement of the foot and ankle are measured using special cameras. The information we get from the gait analysis, in combination with the clinical exam, helps to determine what sort of treatment is necessary. In British Columbia (BC), treatment strategies for children with ITW can include combinations of physical therapy, casts, braces, injections into calf muscles, and/or surgery. Treatment can be time consuming, painful, and costly, yet we do not know if any of these strategies are successful in resolving ITW in the long-term. There is limited data looking at what happens to children who toe walk and we do not know if children with ITW are affected functionally and if there are truly long-term consequences of persistent toe walking.

Your involvement in the study will include a repeat clinical physical therapy exam and three-dimensional gait analysis at the Shriners Gait Lab at Sunny Hill Health Centre for Children. The expected number of participants that will be recruited for the study is 45 children and young adults with a history of ITW.

5. What is the purpose of the study?

The purpose of this study is to determine how many children and young adults are still toe walking at least 10 years following their last visit to the Shriners Gait Lab. We also want to learn whether there is a relationship between the severity of ITW and treatment on long-term functional outcomes. The study will help us learn if activity is limited or if participation is restricted in these children and young adults.

6. Who can participate in this study?

You are eligible to participate if:

- You have or had a diagnosis of ITW
- You were assessed in the Shriners Gait Lab at Sunny Hill Health Centre for Children between the years of 1997-2005
- You received treatment or if you received no treatment at all

7. Who should not participate in this study?

You will not be eligible to participate in this study if, since 2005, you have been diagnosed with one of the following conditions:

- Cerebral palsy
- Spinal cord abnormality
- Muscle disease
- Nerve damage
- Movement disorder
- Autistic spectrum disorder
- Club foot
- One-sided toe walking
- Sudden onset of toe walking

8. What does the study involve?

Overview of the study

The research intervention for this study will include a clinical physical therapy exam, a three-dimensional gait analysis, and a questionnaire. The study will take approximately 60-90 minutes as per a routine three-dimensional gait analysis at the Shriners Gait Lab. The questionnaire takes approximately 15-20 minutes to fill-in and will be completed while the physical therapist prepares the participant for the gait analysis. Participants do not need to answer questions that they are not comfortable answering.

If You Decide to Join This Study: Specific Procedures

If you agree to take part in this study, the procedures and visits you can expect will include the following:

The study will take approximately 60-90 minutes on one day. You will be asked to schedule one appointment at the Shriners Gait Lab at Sunny Hill Health Centre for Children via a telephone call from [redacted]. At this time, any questions you might have about the study will be answered. Once you agree to be a part of the study, your medical records will be accessed only for information related to your toe walking, such as treatment history. When you arrive at the Shriners Gait Lab, a brief clinical exam will be performed by the physical therapist to determine your strength, range of movement, and function around your feet and ankles. This will take 15 minutes. Reflective markers will be placed over your body, mainly on your legs and feet. Wireless EMG stickers will be placed over four muscle groups on your legs. During reflective marker placement, you will be asked to fill in the short questionnaire to determine any activity limitations or participation restrictions as a result of your history of toe walking. Marker placement will take 30 minutes. Gait analysis is a non-invasive part of routine orthopaedic care. The gait analysis uses a 12-camera Motion Analysis system to record the three-dimensional positions of reflective markers. You will be asked to walk at a comfortable pace several times while cameras record your movement. This will take 15-30 minutes. Your face will be blurred in the video recordings and you will not be identifiable. The video recordings will be stored in a

secure computer server only accessible by the research team at the Gait Lab. The video recordings will be used to help analyze how you walk for the sole purpose of this study. All video recordings will be removed from the camera once data analysis for the study has been completed. You will also be asked to walk across a mat that shows areas of high and low pressure under your feet while you walk. This will take 15 minutes.

The study assessment will be set for a time that works well for your family. Parents can stay for the whole assessment.

9. What are my responsibilities?

Participants should wear tight-fitting shorts and a tank top or a T-shirt so it is easy to apply the reflective markers and EMG stickers.

10. What are the possible harms and discomforts?

There are no known risks to you for participating in this project. All tests performed for this study are part of the routine care plan you have experienced in the past at the Shriners Gait Lab. You may experience some mild irritation at the site of the markers due to the tape that is used to attach the markers to your skin. If toe walking or some related physical characteristic is still occurring, the physical therapist will recommend you contact your family physician to discuss any concerns.

11. What are the potential benefits of participating?

There are no proven direct benefits to you for participating in this study.

The results of this study may help future individuals who are diagnosed with idiopathic toe walking. The results may potentially reduce or prevent inherent costs in terms of discomfort, pain, time, and specific expenses for children and their families.

12. What happens if I decide to withdraw my consent to participate?

You may withdraw from this study at any time without giving reasons. If you choose to enter the study and then decide to withdraw at a later time, you have the right to request the withdrawal of your information collected during the study. This request will be respected to the extent possible. Please note however that there may be exceptions where the data will not be able to be withdrawn for example where the data is no longer identifiable (meaning it cannot be linked in any way back to your identity) or where the data has been merged with other data. If you would like to request the withdrawal of your data, please let [redacted] know.

13. Can I be asked to leave the study?

If you are not able to follow the requirements of the study or for any other reason, the principal investigator may withdraw you from the study.

14. How will my taking part in this study be kept confidential?

Your confidentiality will be respected. However, research records and health or other source records identifying you may be inspected in the presence of the Investigator for the purpose of monitoring the research. No information or records that disclose your identity will be published without your consent, nor will any information or records that disclose your identity be removed or released without your consent unless required by law.

You will be assigned a unique study number as a participant in this study. This number will not include any personal information that could identify you (e.g., it will not include your Personal Health Number, SIN, or your initials, etc.). Only this number will be used on any research-related information collected about you during the course of this study, so that your identity will be kept confidential. Information that contains your identity will remain only with the Principal Investigator and/or designate. The list that matches your name to the unique study number that is used on your research-related information will not be removed or released without your consent unless required by law.

Your rights to privacy are legally protected by federal and provincial laws that require safeguards to insure that your privacy is respected. You also have the legal right of access to the information about you that has been provided to the sponsor and, if need be, an opportunity to correct any errors in this information. Further details about these laws are available on request to the Principal Investigator.

15. What happens if something goes wrong?

By signing this form, you do not give up any of your legal rights and you do not release the investigators, participating institutions, or anyone else from their legal and professional duties.

16. What will the study cost me?

There are no parking costs at Sunny Hill Health Centre for Children.

Remuneration

You will not receive any payment for participation in this study. The physical therapist will provide you with a letter outlining your volunteer hours for use for high school volunteer requirements.

17. Who do I contact if I have questions about the study during my participation?

If you have any questions or desire further information about this study before or during participation, or if you experience any adverse effects, you can contact [redacted]

18. Who do I contact if I have any questions or concerns about my rights as a participant?

If you have any concerns or complaints about your rights as a research participant and/or your experiences while participating in this study, contact the Research Participant Complaint Line in the University of British Columbia Office of Research Ethics by e-mail at RSIL@ors.ubc.ca or by phone at 604-822-8598 (Toll Free: 1-877-822-8598).

19. After the study is finished

Once the study is completed, we will send participants a summary of the findings. This may be up to one year after you agree to participate in the study.

Future Contact

If you wish to be contacted by the Principal Investigator for future studies, please indicate so below by ticking the appropriate box:

☐ Yes, I would like to be contacted for futures studies

☐ No, please do not contact me for future studies

Participant Consent

My signature on this consent form means:

- I have read and understood the information in this consent form.
- I have had enough time to think about the information provided.
- I have been able to ask for advice if needed.
- I have been able to ask questions and have had satisfactory responses to my questions.
- I understand that all of the information collected will be kept confidential and that the results will only be used for scientific purposes.
- I understand that my participation in this study is voluntary.
- I understand that I am completely free at any time to refuse to participate or to withdraw from this study at any time, and that this will not change the quality of care that I receive.
- I authorize access to my health records as described in this consent form.

- I understand that I am not waiving any of my legal rights as a result of signing this consent form.
- I understand that there is no guarantee that this study will provide any benefits to me.

The parent(s)/guardian(s)/substitute decision-maker (legally authorized representative) and the investigator are satisfied that the information contained in this consent form was explained to the child/participant to the extent that he/she is able to understand it, that all questions have been answered, and that the child/participant assents to participating in the research.

I will receive a signed copy of this consent form for my own records.

I consent to participate in this study.

_____	_____	_____
Participant's or Substitute Decision-Maker's Signature	Printed name	Date

_____	_____	_____	_____
Signature of Person Obtaining Consent	Printed name	Study Role	Date

Appendix C: Adolescent Assent Form

ADOLESCENT INFORMATION AND ASSENT FORM
Long-term Follow-up of Idiopathic Toe Walking

Principal Investigator: [redacted]

Co-Investigators [redacted]

Invitation

I am being invited to be part of a research study. A research study tries to find better ways to help adolescents like me. The following pages explain the study so that I can decide if I want to take part or not. It is up to me if I want to be in this study. No one will make me be part of the study. Even if I agree now to be part of the study, I can change my mind later. No one will be mad at me if I choose not to be part of this study.

Do I Have to be in This Study?

If I want to participate in this study, I will be asked to sign this form. My parent/guardian will need to sign a consent form before I am enrolled in the study; but I do not have to participate even if they sign the consent form. The researchers will not enroll me into the study unless I agree to do so.

Why Are We Doing This Study?

I had a condition called idiopathic toe walking. This condition affects many other children. Some children walk on their toes when they are really little, and others keep walking on their toes until they grow older. This study to trying to find out how many children that were seen at the Shriners Gait Lab at Sunny Hill Health Centre for Children are still toe walking now that they are older children. This study will help us learn what kinds of things help or get in the way of everyday activities for children with idiopathic toe walking.

Why Are You Inviting Me to be in This Study?

I am being invited to be in this study to see what changes take place in idiopathic toe walking after at least 10 years and to see if toe walking makes it difficult to do any of the activities that I like to do. We are expecting 45 participants to take part in this study.

What Will Happen in This Study?

If I agree to be in this study, I will go to the Gait Lab at Sunny Hill Health Centre for Children for a follow-up visit where the physical therapist will look at how strong my muscles are and how much movement my muscles have around my feet and ankles. The physical therapist will attach some reflective stickers onto my feet and legs and ask me to fill in one questionnaire about how easy or hard it is to do the activities I like. The stickers will help cameras to take my picture more easily while I am walking. I will be asked to walk at a comfortable speed while the cameras take my picture. I will be identifiable in the camera recordings and the pictures will be stored in a secure computer server only accessible by the research team at the Gait Lab. The pictures will be used to help analyze how I walk. I will also be asked to walk over a special mat that can take pictures of the bottom of my feet. The physical therapist is going to look into my hospital chart to get some information about what kind of treatment I had for my toe walking. I only have to come on one day. This visit will take approximately 1-1.5 hours.

Who is Doing This Study?

[redacted] and her research team from Sunny Hill Health Centre for Children will be doing this study. They will answer any questions I have about the study. I can also call them at [redacted] if I am having any problems.

Can Anything Bad Happen to Me?

The researcher does not think there are any bad things about doing this study. I may feel a little bit sticky or my skin may be a little bit red where the physical therapist puts the stickers on my skin. This should go away quickly and the sticky feeling washes off with water. If I am still toe walking or if something related to toe walking is still occurring, the physical therapist will recommend I contact my family physician to discuss my concerns.

Who Will Know I Am in the Study?

Only the people involved in the study will know I am in it. When the study is finished, the researchers will write a report about what was learned. This report will not say my name or that I was in the study. My parents and I do not have to tell anyone I am in the study if we do not want to.

What Will the Study Cost Me?

There are no parking costs at Sunny Hill Health Centre for Children.

Remuneration

I will not receive any payment for participation in this study. The physical therapist will provide me with a letter outlining my volunteer hours for use for high school volunteer requirements.

When Do I Have to Decide?

I have as much time as I want to decide to be part of the study. I have also been asked to discuss my decision with my parents.

Who do I Contact if I Have Questions about the Study during My Participation?

If I have any questions or desire further information about this study before or during participation, or if I experience any adverse effects, I can contact [redacted]

Who do I Contact if I Have Any Questions or Concerns about My Rights as a Participant?

If I have any concerns or complaints about my rights as a research participant and/or my experiences while participating in this study, I should contact the Research Participant Complaint Line in the University of British Columbia Office of Research Ethics by e-mail at RSIL@ors.ubc.ca or by phone at 604-822-8598 (Toll Free: 1-877-822-8598).

Future Studies

There is a chance that during or after this study the study team will find other questions needing answers that require future studies. If I am willing to hear about these future studies I will mark the "yes" box. This does not mean that I will have to take part in a new study, just that the study team will let me know about it. If I do not want to be contacted about new studies I will mark the "no" box."

Are you willing to be contacted by the researchers for future studies?

YES □

NO □

Assent to Participate

My signature on this assent form means:

- I have read and understood this adolescent information and assent form.
- I have had enough time to consider the information provided and to ask for advice if necessary.
- I have had the opportunity to ask questions and have had acceptable answers to my questions.
- I understand that all of the information collected will be kept confidential and that the results will only be used for scientific objectives.
- I understand that my participation in this study is voluntary and that I am completely free to refuse to participate or to withdraw from this study at any time without changing the quality of care that I receive.
- I understand that I can continue to ask questions, at any time, regarding my participation in the study.
- I understand that if I put my name at the end of this form, it means that I agree to be in this study.

I will receive a signed copy of this assent form for my own records.

I agree to participate in this study.

_____	_____	_____
Participant's Signature	Printed name	Date
_____	_____	_____
Name of Person Who Obtained Assent	Signature	Date

APPENDIX D: Reliability Analysis for Sagittal Ankle Angles

Comparison of means, standard deviation (SD), and standard error of measurement (SEM) for master's candidate (PT1) and physical therapist at baseline (PT2)

Gait Analysis Variable	**Mean**		**SD**		**SEM**	
	PT1	**PT2**	**PT1**	**PT2**	**PT1**	**PT2**
Ankle angle at initial contact	-3.8	-2.7	1.7	1	1.3	0.6
Peak ankle DF, stance	9.1	10.3	1.4	1.8	0.9	0.7
Peak ankle DF, swing	1.0	1.6	2.1	2.6	1.1	0.9
Peak knee extension, stance	-1.9	-1.8	4.0	2.8	1.1	1.1
Peak knee flexion, swing	59.1	60.2	3.4	2.7	1.3	1.5

Appendix E: Pediatric Outcomes Data Collection Instrument

Adolescent (self-reported)

Outcomes Questionnaire

Developed by:
American Academy of Orthopaedic Surgeons®
Pediatric Orthopaedic Society of North America
American Academy of Pediatrics
Shriner's Hospitals
To be completed by adolescents 11 – 18 years old.
Based on the Version 2.0 Pediatrics–Parent/ Adolescent Outcomes Instrument
Also commonly referred to as the PODCI ("Pediatric Outcomes Data Collection Instrument")
Revised, renumbered, reformatted August 2005

Some kind of problems can make it hard to do many activities, such as eating, bathing, school work, and playing with friends. We would like to find out how you are doing. (Choose one response per line.)

During the **last week,** was it easy or hard for you to**:**

1. Lift heavy books?

1 - Easy
2 - A little hard
3 - Very hard
4 - Can't do at all

2. Pour a half gallon of milk?

3. Open a jar that has been opened before?

4. Use a fork and spoon?

5. Comb your hair?

6. Button buttons?

7. Put on your coat?

8. Write with a pencil?

9. On average, **over the last 12 months,** how often did you miss school (camp, etc.) because of your health?

- *Rarely*
- *Once a month*
- *Two or three times a month*
- *Once a week*
- *More than once a week*
- *Do not attend school, etc.*

During the **last week,** how happy have you been with: (Choose one response per line.)

10. How you look?

1 - Very happy
2 – Somewhat happy
3 - Not sure

4 – Somewhat unhappy
5 - Very unhappy

11. Your body?

12. What clothes or shoes you can wear?

13. Your ability to do the same things your friends do?

14. Your health in general?

During the **last week,** how much of the time: (Choose one response per line.)

15. Did you feel sick and tired?

1 - Most of the time
2 - Some of the time
3 - A little of the time
4 - None of the time

16. Were you full of pep and energy?

17. Did pain or discomfort interfere with your activities?

During the **last week,** has it been easy or hard for you to: (Choose one response per line.)

18. Run short distances?

1 - Easy
2 - A little hard
3 - Very hard
4 - Can't do at all

19. Bicycle or tricycle?

20. Climb three flights of stairs?

21. Climb one flight of stairs?

22. Walk more than a mile?

23. Walk three blocks?

24. Walk one block?

25. Get on and off a bus?

26. How often do you need help from another person for walking and climbing? (Choose one response.)
1 - Never 2 - Sometimes 3 - About half the time 4 - Often 5 - All the time

27. How often do you use assistive devices (such as braces, crutches, or wheelchair) for walking and climbing?
(Choose one response.)
1 - Never 2 - Sometimes 3 - About half the time 4 - Often 5 - All the time

During the **last week**, has it been easy or hard for you to: (Choose one response per line.)

28. Stand while washing your hands and face at a sink?

1 - Easy
2 - A little hard

3 - Very hard
4 - Can't do at all

29. Sit in a regular chair without holding on?

30. Get on and off a toilet or chair?

31. Get in and out of bed?

32. Turn door knobs?

33. Bend over from a standing position and pick up something off the floor?

34. How often do you need help from another person for sitting and standing? (Choose one response.)

1 - Never *2 - Sometimes* *3 - About half the time* *4 - Often* *5 - All the time*

35. How often do you use assistive devices (such as braces, crutches, or wheelchair) for sitting and standing?
(Choose one response.)

1 - Never *2 - Sometimes* *3 - About half the time* *4 - Often* *5 - All the time*

36. Can you participate in **recreational outdoor activities** with other kids the same age? (For example: bicycling, skating, hiking, jogging) (Choose one response.)

1 - Yes, easily *2 - Yes, but a little hard* *3 - Yes, but very hard* *4 - No*

If you answered "no" to Question 36 above, was your activity limited by: (Choose all that apply.)

37. *Pain?*

38. *General health?*

39. *Doctor or parent instructions?*

40. *Fear the other kids won't like you?*

41. *Dislike of recreational outdoor activities?*

42. *Activity not in season?*

43. Can you participate in **pickup games or sports** with other kids the same age? (For example: tag, dodge ball, basketball, softball, soccer, catch, jump rope, touch football, hop scotch)
(Choose one response.)

1 - Yes, easily *2 - Yes, but a little hard* *3 - Yes, but very hard* *4 – No*

If you answered "no" to Question 43 above, was your activity limited by: (Choose all that apply.)

44. *Pain?*

45. *General health?*

46. *Doctor or parent instructions?*

47. *Fear the other kids won't like you?*

48. *Dislike of pickup games or sports?*

49. *Activity not in season?*

50. Can you participate in **competitive level sports** with other kids the same age? (For example: hockey, basketball, soccer, football, baseball, swimming, running [track or cross country], gymnastics, or dance) (Choose one response.)

1 - Yes, easily 2 - Yes, but a little hard 3 - Yes, but very hard 4 – No

If you answered "no" to Question 50 above, was your child's activity limited by: (Choose all that apply.)

51. *Pain?*
52. *General health?*
53. *Doctor or parent instructions?*
54. *Fear the other kids won't like you?*
55. *Dislike of competitive level sports?*
56. *Activity not in season?*

57. How often in the **last week** did you get together and do things with friends? (Choose one response.)

1 - Often 2 - Sometimes 3 - Never or rarely

If you answered "sometimes" or "never or rarely" to Question 57 above, was your activity limited by: (Choose all that apply.)

58. *Pain?*
59. *General health?*
60. *Doctor or parent instructions?*
61. *Fear the other kids won't like you?*
62. *Friends not around?*

63. How often in the **last week** did you participate in **gym/recess**? (Choose one response.)

1 - Often 2 - Sometimes 3 - Never or rarely 4 - No gym or recess

If you answered "sometimes" or "never or rarely" to Question 63 above, was your activity limited by: (Choose all that apply.)

64. *Pain?*
65. *General health?*
66. *Doctor or parent instructions?*
67. *Fear the other kids won't like you?*
68. *Dislike of gym/recess?*
69. *School not in session?*
70. *I don't attend school?*

71. Is it easy or hard for you to make friends with kids your own age? (Choose one response.)

1 - Usually easy 2 - Sometimes easy 3 - Sometimes hard 4 - Usually hard

72. How much pain have you had during the **last week**? (Choose one response.)

1 - None 2 - Very mild 3 - Mild 4 - Moderate 5 - Severe 6 - Very severe

73. During the **last week,** how much did pain interfere with your normal activities (including at home, outside of the home, and at school)? (Choose one response.)

1 - Not at all 2 - A little bit 3 - Moderately 4 - Quite a bit 5 - Extremely

What expectations do you have for your treatment?

As a result of my treatment, I expect: (Choose one response per line.)

74. To have pain relief.

1 - Definitely yes
2 - Probably yes
3 - Not sure

4 - Probably not
5 - Definitely not

75. To look better.
76. To feel better about myself.
77. To sleep more comfortably.
78. To be able to do activities at home.
79. To be able to do more at school.
80. To be able to do more play or recreational activities (biking, walking, doing things with friends).
81. To be able to do more sports.
82. To be free from pain or disability as an adult.

83. If you had to spend the rest of your life with your bone and muscle condition **as it is right now,** how would you feel about it? (Choose one response.)

1 - Very satisfied
2 – Somewhat satisfied
3 - Neutral
4 – Somewhat dissatisfied
5 - Very dissatisfied

Comments:

Appendix F: Medical Outcomes Study 36-Item Short Form Survey

Your Health and Well-Being

This survey asks for your views about your health. This information will help keep track of how you feel and how well you are able to do your usual activities. *Thank you for completing this survey!*

For each of the following questions, please mark an ☒ in the one box that best describes your answer.

1. In general, would you say your health is:

Excellent	Very good	Good	Fair	Poor
☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

2. <u>Compared to one year ago</u>, how would you rate your health in general <u>now</u>?

Much better now than one year ago	Somewhat better now than one year ago	About the same as one year ago	Somewhat worse now than one year ago	Much worse now than one year ago
☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

3. The following questions are about activities you might do during a typical day. Does <u>your health now limit you</u> in these activities? If so, how much?

	Yes, limited a lot	Yes, limited a little	No, not limited at all
a	<u>Vigorous activities</u>, such as running, lifting heavy objects, participating in strenuous sports	☐ 1	☐ 2	☐ 3
b	<u>Moderate activities</u>, such as moving a table, pushing a vacuum cleaner, bowling, or playing golf.....	☐ 1	☐ 2	☐ 3
c	Lifting or carrying groceries	☐ 1	☐ 2	☐ 3
d	Climbing <u>several</u> flights of stairs	☐ 1	☐ 2	☐ 3
e	Climbing <u>one</u> flight of stairs	☐ 1	☐ 2	☐ 3
f	Bending, kneeling, or stooping	☐ 1	☐ 2	☐ 3
g	Walking <u>more than a kilometre</u>	☐ 1	☐ 2	☐ 3
h	Walking <u>several hundred metres</u>	☐ 1	☐ 2	☐ 3
i	Walking <u>one hundred metres</u>	☐ 1	☐ 2	☐ 3
j	Bathing or dressing yourself	☐ 1	☐ 2	☐ 3

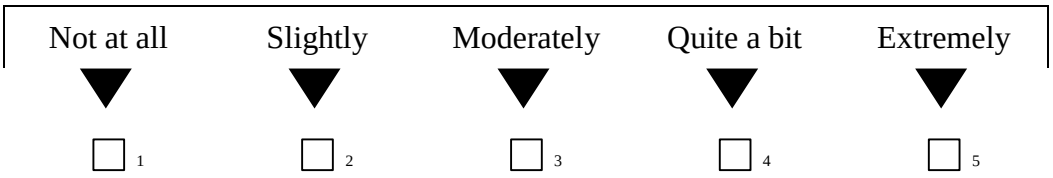
4. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of your physical health?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a Cut down on the amount of time you spent on work or other activities	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
b Accomplished less than you would like	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
c Were limited in the kind of work or other activities	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
d Had difficulty performing the work or other activities (for example, it took extra effort)	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

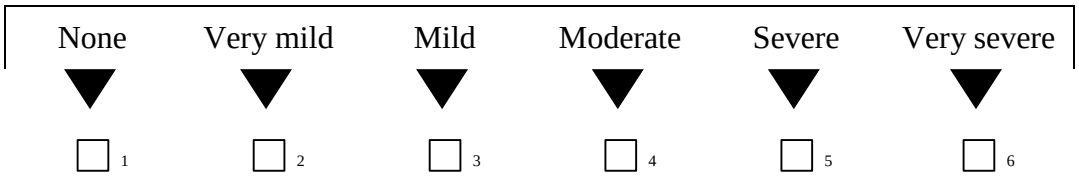
5. During the past 4 weeks, how much of the time have you had any of the following problems with your work or other regular daily activities as a result of any emotional problems (such as feeling depressed or anxious)?

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a Cut down on the amount of time you spent on work or other activities	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
b Accomplished less than you would like	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
c Did work or other activities less carefully than usual	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

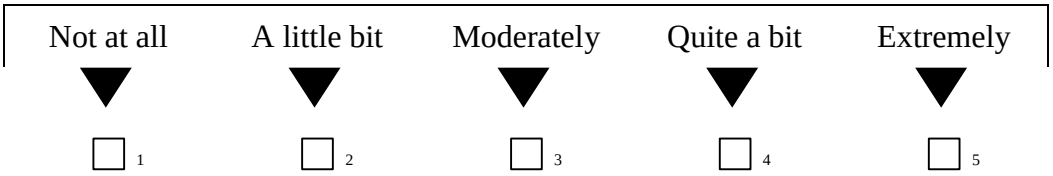
6. During the <u>past 4 weeks</u>, to what extent has your physical health or emotional problems interfered with your normal social activities with family, friends, neighbours, or groups?



7. How much <u>bodily</u> pain have you had during the <u>past 4 weeks</u>?



8. During the <u>past 4 weeks</u>, how much did <u>pain</u> interfere with your normal work (including both work outside the home and housework)?



9. These questions are about how you feel and how things have been with you <u>during the past 4 weeks</u>. For each question, please give the one answer that comes closest to the way you have been feeling. How much of the time during the <u>past 4 weeks</u>…

	All of the time	Most of the time	Some of the time	A little of the time	None of the time
a Did you feel full of life?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
b Have you been very nervous?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
c Have you felt so down in the dumps that nothing could cheer you up?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
d Have you felt calm and peaceful?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
e Did you have a lot of energy?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
f Have you felt downhearted and depressed?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
g Did you feel worn out?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
h Have you been happy?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
i Did you feel tired?	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

10. During the <u>past 4 weeks</u>, how much of the time has your <u>physical health or emotional problems</u> interfered with your social activities (like visiting with friends, relatives, etc.)?

All of the time	Most of the time	Some of the time	A little of the time	None of the time
☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

11. How TRUE or FALSE is <u>each</u> of the following statements for you?

	Definitely true	Mostly true	Don't know	Mostly false	Definitely false
a I seem to get sick a little easier than other people	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
b I am as healthy as anybody I know	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
c I expect my health to get worse	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5
d My health is excellent	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5

Thank you for completing these questions!

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License Number: QM030263

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Licensee Address: [redacted]

Approved Purpose: Long-term follow-up of children with idiopathic toe walking

Study Type: Non-commercial academic research and/or thesis – Unfunded Student
Data Collection Method: Paper

Therapeutic Area: Wellness & Lifestyle

Royalty Fee: None, because this License is granted in support of the non-commercial Approved Purpose

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B. Appendices: Capitalized terms used in this Agreement shall have the meanings assigned to them in Appendix A and Appendix B. The appendices attached hereto are incorporated into and made a part of this Agreement for all purposes.

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EXECUTED by the duly authorized representatives as set forth below.

OptumInsight Life Sciences, Inc. [redacted]

LICENSE AGREEMENT - DETAILS

Licensee: [redacted]

License Number: [redacted]

Study Term: 06/03/15 to 06/02/16

Amendment to: N/A

Approved Purpose: Long-term follow-up of children with idiopathic toe walking

Licensed Surveys (Modes) and Services: Item Description

ES0220 SF-36v2, Standard Recall 1 Paper

Approved Languages: Canada (English)

IS0220 SF-36v2 Interview Script, Std 1 Interview Script

Approved Languages: Canada (English)

SS075 Scoring Software v4.5 1

SS080 SS v4.5 Key: SF-36v2 100

EM125 SF-36v2 User's Manual 3rd Ed. 1

Approved Languages: United States (English)

TOTAL FEES: 0.00 USD