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Self-rated walking disability and dynamic ankle joint stiffness in children and adolescents with Juvenile Idiopathic Arthritis receiving intraarticular corticosteroid joint injections of the foot

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ABSTRACT

Background: Children and adolescents with Juvenile Idiopathic Arthritis (JIA) exhibit deviations in ankle dynamic joint stiffness (DJS, or moment-angle relationship) compared to healthy peers, but the relationship between ankle DJS and self-reported walking impairments has not been studied. This secondary analysis aimed to investigate the relationship between ankle DJS and self-reported walking disability in juveniles with JIA, and to determine whether intraarticular corticosteroid foot injections (IACI) were associated with long term changes in ankle DJS.

Research questions: Is ankle DJS altered in children with JIA reporting walking difficulties compared to children with JIA reporting no walking difficulties? Are IACIs associated with persistent alterations in ankle DJS?

Methods: Gait dynamics (DJS), foot pain, and foot-related disability were assessed in 33 children with JIA before intraarticular corticoid foot injection (IACI), and three months after IACI. Using self-reported walking capacity scores, children were classified as either having no walking difficulties (ND) or having walking difficulties (WD). Inferential statistics were used to compare demographics, pain, impairment scores, and ankle DJS between the groups.

Results: Before treatment, in the WD group, ankle DJS was significantly decreased both in the early rising phase (ERP = 0.03±0.02 vs. 0.05±0.02 Nm(kg*deg)$^{-1}$) and late rising phase (LRP = 0.11±0.06 vs. 0.24+0.22 Nm (kg*deg)$^{-1}$) compared to the ND group. At three months, the ERP was still significantly decreased in the WD group (ERP = 0.03±0.01 vs. 0.05±0.03 Nm(kg*deg)$^{-1}$).

Significance: Among children and adolescents with JIA who reported walking difficulties prior to IACIs, alterations in DJS in early stance phase (decreased ERP) remained three months after IACI suggesting persistent gait adaptations, possibly related to pain. Pre-treatment gait analysis may aid in identifying children who will not have long term benefit from IACIs in terms of improved gait, and therefore, may be informed and have the choice to be spared the risk of side effects associated with this treatment.

1. Introduction

Juvenile idiopathic arthritis (JIA) is the most common rheumatic disease in childhood and refers to all forms of arthritis that are present before 16 years of age, persist for more than six weeks, and are of unknown etiology [1]. JIA is subclassified by the pattern of symptoms into oligoarthritis (involving one to four joints); polyarthritis (more than four joints); systemic arthritis (including a range of systemic features), psoriatic arthritis, enthesitis-related arthritis (mostly HLA B-27 related disease), and an undefinable group referred to as "other arthritis" [2]. The ankle joint is the second most commonly affected joint after the knee, but JIA may also involve other foot joints causing pain,

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deformity, and gait alterations [3,4]. The goal of pharmacotherapy in JIA is to reduce synovitis and to hopefully achieve remission in order to prevent joint and systemic damage [5]. Medications to manage JIA are potentially toxic and intraarticular corticosteroid injections may help to reduce or eliminate the need for these potent drugs. Indications for IACI include resolution of synovitis not currently controlled with systemic medications and the goal of inducing remission [5].

The Child Health Assessment Questionnaire (CHAQ) [6] and Juvenile Arthritis Foot Disability Index (JAFI) [7] are commonly used self-reported outcome measures designed to evaluate pain and disability in children with JIA and to describe the impact of JIA on function.

Three-dimensional gait analysis (3D-GA) provides more detailed and quantitative data regarding gait variations [8,9]. In children with polyarticular JIA, 3D-GA demonstrates that these children walk with increased hip and knee flexion and reduced ankle plantarflexion [9]. Reduced walking speed and increased double limb support time are common gait alterations recognized in patients with JIA who have ankle involvement [10].

The dynamics of single joints can be further analyzed by combining kinematic and kinetic data, such as the ankle moment-angle relationship or dynamic joint stiffness (DJS) [11]. DJS offers an non-invasive analysis of two dynamic joint functions; the spring-like behavior of the joint and mechanical energy exchanges during the movement [12]. In the tibiotalar joint, this curve forms a relatively simple loop-shaped contour during the stance phase [11,13], where four sequential linear phases can be discerned: 1) the first descending phase (FDP), 2) the early rising phase (ERP), 3) the late rising phase (LRP) and 4) the descending phase, (DP) [12]. Temporally the ERP, LRP and DP correspond approximately to the ankle, forefoot and toe rockers described by Perry and Burnfield [14]. DJS in the ankle has been described in eight children with JIA [15]. In that study, the ERP was significantly lower than in healthy controls, while the LRP and DP were not. The lower ERP was ascribed to a lower ground reaction force (GRF) and/or slower advancement of the center of pressure (COP) [15]. However, the relationship between ankle DJS and walking disability was not explored.

The present study is based on Esbjornsson et al.'s [16] observation that children with JIA who reported severe walking difficulties before foot IACIs demonstrated less improvement in gait dynamics, as measured by the gait deviation index (GDI) and lower ankle /hip power ratio before, as well as three months after IACI treatment compared to children with JIA reporting no pre-treatment walking difficulties. In that study all participants received foot and ankle IACIs but the biomechanics of the ankle joint was not analyzed in detail. This study aimed to investigate whether ankle DJS varied depending on self-rated walking disability in children and adolescents with JIA in order to determine whether a subgroup of children with JIA and gait impairments might respond less well to foot IACI therapy in terms of persisting gait changes.

2. Materials and methods

The regional ethical board in Stockholm, Sweden approved this study. All participants (and/or parents of children) provided informed written consent. The study design and methods have previously been described [16].

This secondary analysis examines data from 33 juveniles with JIA who were consecutively recruited from Astrid Lindgren's Children's Hospital in Stockholm, Sweden. The inclusion criteria were: 1) active arthritis in one or both feet, 2) scheduled for IACIs, 3) age between 5 years and 18 years, 4) typically developing, 5) able to understand written and spoken Swedish and completion of the baseline and three months assessments (questionnaires and 3D-GA). Children who had received major lower limb surgery or IACIs less than 4 weeks before baseline assessments were excluded. Sixty-four percent of the participants were diagnosed with polyarthritis and 58% received IACIs in the knee and/or hip joint. Seventy-three percent of the children were taking non-steroid anti-inflammatory drugs, 42% methotrexate, and 33% biologic agents. Participants received physiotherapy as needed. Injections were given under general anesthesia and fluoroscopy with contrast was used to verify correct needle placement. Eight children also participated in an earlier study on ankle DJS in children with JIA, idiopathic toe-walkers and healthy controls [15].

Children were examined before treatment, at three weeks, and three months after IACIs. At each assessment, children rated their pain during walking, underwent a clinical joint examination by an experienced physical therapist (ACE), and participated in a 3D-GA. Self-reported foot pain was estimated using either a Visual Analog Scale (VAS) (scale ranged from 1 to 100), or a faces pain scale, depending on the child's age and comprehension ability. The clinical examination consisted of an assessment of 1) capsular swelling or effusion in the knee, ankle, and forefoot, 2) presence of tenderness and pain in the hip, knee, ankle, mid/hindfoot, and forefoot, 3) loss of joint motion (same 5 joints as 2) and dichotomized as either present or absent. For each joint investigated, the figures were added to calculate a joint score [17], with a maximum value of 26 (including both extremities).

Children completed the Juvenile Foot Disability Index (JAFI) [7], a measure of foot dysfunction, and the Child Health Assessment Questionnaire (CHAQ) [6] at baseline and at three months. The CHAQ and JAFI are commonly used valid and reliable self-reported outcome measures designed to evaluate pain and disability in children with JIA. For children younger than 10 years of age (n = 14) a proxy version of the JAFI and CHAQ was completed by the same parent at baseline and three months. When completing the JAFI, the most involved foot served as the reference foot. The CHAQ assessed function in eight areas of daily living. Each item uses 0–3 response set (0 =" without difficulty" to 3 = "impossible to do"). Children were dichotomized into two groups based on their baseline CHAQ walking dimension score (no difficulty (ND) = score 0 or walking difficulty (WD) = score 1 or 2). No child scored 3 =" impossible to do".

Gait data was collected before and after IACI using an eight-camera motion analysis system (Vicon MX40, Oxford, UK), with two force plates (Kistler Type 9281C, Winterthur, Switzerland). Data were analyzed with the Vicon Plug-In-Gait model. An experienced physiotherapist (ACE) placed markers and conducted the gait analyses on all children. Relevant data concerning ankle kinematics, moment, and gait events as well as marker trajectories, ground reaction force and center of pressure data were analyzed using Matlab (MathWorks, Inc., Natick, USA). Linear interpolation was used to obtain data points for joint angles and moments at every 2% of stride duration. Ankle joint moment was computed by inverse dynamics, using subjects' measurements and anthropometric properties [18] and normalized to body weight. The ankle moment (stance phase only) was plotted as a function of the corresponding ankle angle (moment-angle loop) in the sagittal plane. Similar to previous reports [13,15], the moment-angle loop forms a counter-clockwise transverse path, comprising three distinctive quasi-linear phases corresponding to the events of the ankle joint during the stance phase (Fig. 1A). In the ERP, the ankle passively dorsiflexes through the rotation of the tibia over the stationary foot. The LRP starts from heel rise and ends at maximal ankle dorsiflexion. The DP subsequently starts and is completed when the foot is off the ground. Based on the literature [13], a threshold value is applied to trim off the turning points to avoid non-linearity. The determination scheme of each subphase comprising the moment-ankle loop has been described in detail in previous studies [13,15]. The high goodness-of-fit of linear regression in each sub-phase indicated that the linear phase was successfully identified. For each sub-phase, ankle DJS was quantified as the slope of the moment-angle curve using a linear regression line, minimizing the least square distance between the experimental data points and the line. Due to the small number of data points, FDP was excluded in the analysis. An increase or a decrease in the slope of the different phases will be referred to as an increase or decrease of e.g. the ERP.

The areas subtended by the rising and the descending components

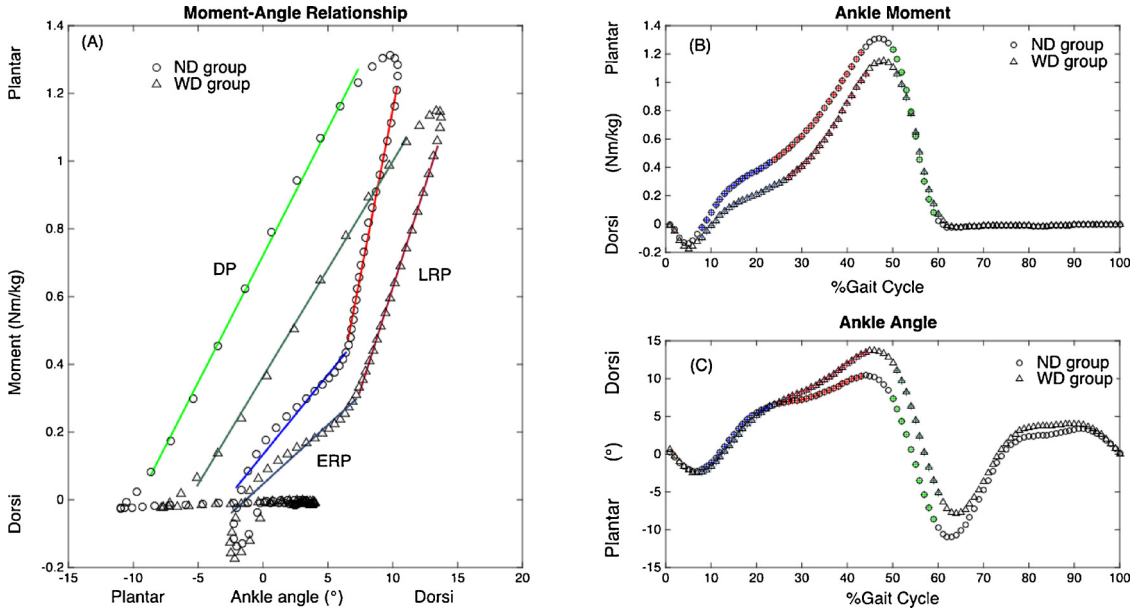

Fig. 1. (A) Moment-angle relationship showing averaged moment-angle loops from 20 children with self-reported walking difficulties "WD" and 13 children reporting no walking difficulties "ND" before treatment. Dynamic ankle joint stiffness was calculated as the slope of the linear regression line of ankle joint moment plotted as a function of ankle joint angle. Illustrated are the three sub-phases within the stance phase; early rising phase "ERP" (blue), late rising phase "LRP" (red), and descending phase "DP" (green). (B) Ankle Moment showing averaged ankle moments normalized to gait cycle length in the WD and ND groups. The internal plantarflexion moment is lower in the WD group during the ankle and forefoot rockers, (C) Ankle angle variations during the normalized gait cycle. The ankle dorsiflexion angle is larger in the WD group during the later part of the stance phase. (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

of the loop, correspond to the work absorbed and the work produced, respectively (Fig. 2). The area within the loop represents the net work produced by the ankle joint. Data from the most involved foot (same as for completion of JAFI score) was used for analysis. In the case of bilateral foot involvement, data from the most affected foot, as indicated by the child, was used in the analysis.

2.1. Statistical analysis

Statistical analyses were performed using Statistical Package for Social Sciences, version23 (SPSS Inc., Chicago, IL USA). Demographic and disease characteristics were described using either means and standard deviations (SD) or medians and ranges. Children were divided into two groups according to their baseline CHAQ walking dimension scores. For group comparisons, the Student's *t*-test was used for demographic (age, weight, length, BMI, disease duration) and gait parameters (DJS, work, walking speed), while the Mann-Whitney U test was used for ordinal variables and variables with non-parametric distributions (Joint Score, CHAQ, JAFI but also LRP due to skewed distribution). Spearman rank correlation coefficients were used to assess associations between variables. A p-value < 0.05 was considered statistically significant, otherwise it was referred to as not significant (n.s.).

3. Results

3.1. Demographics, pain, health and disability

Thirteen children scored 0 in the CHAQ walking dimension and are referred to as the "ND" group (no walking disability), while the twenty children who scored 1 (n = 14) or 2 (n = 6) are referred to as the "WD" group (with walking disability). Group comparisons of demographics, pain and questionnaire scores are shown in Table 1. Most subjects were females. There were no differences in age, height or body weight between the ND and WD groups. BMI and pretreatment pain were significantly higher in the WD group. Total CHAQ scores as well as all three JAFI scores were significantly worse **in** children reporting walking disability compared to those not reporting walking disability (See Table 1).

3.2. Ankle dynamic joint stiffness, work and non-dimensional walking speed

When comparing the ND and WD groups before IAC treatment, the ERP and the LRP were significantly decreased ($p < 0.05$) in the WD group, (Table 2, Fig. 1a). There were no significant differences concerning the descending phase (DP), work, or walking speed (Table 2).

At three months, the ERP was still significantly lower ($p < 0.05$) in the WD group compared to the ND group (ERP = 0.03 ± 0.01 vs. 0.05 ± 0.03 Nm(kg*deg)$^{-1}$), but no significant differences were found concerning LRP, DP, work, or non-dimensional walking speed (Table 2).

4. Discussion

Esbjornsson et al. [16] observed that children with JIA reporting severe walking problems had worse gait disability index (GDI) scores and lower ankle/hip power ratio before, as well as three months after IAC treatment, while children reporting no walking problems had a GDI similar to healthy controls. In this study, we aimed to identify the differences in ankle dynamic joint stiffness (DJS) based on the self-reported walking difficulty in children with JIA and to determine whether self-reported walking difficulty could inform the use of foot IACI. We found significant alterations in ankle DJS in early stance phase.

Before IAC treatment, the ERP and LRP values were significantly lower in the group of children with JIA reporting walking difficulties (WD) than in the group with no difficulties (ND). Temporally these changes in ERP and LRP (Fig. 1) occur during the ankle and forefoot rockers described by Perry and Burnfield [14]. Thus, at baseline, the WD group showed a reduced dorsiflexion moment during the ankle and forefoot rockers compared to the ND group. The ankle dorsiflexion angle was also increased during the forefoot rocker in the WD group. After three months, only ERP was reduced in the WD group, suggesting

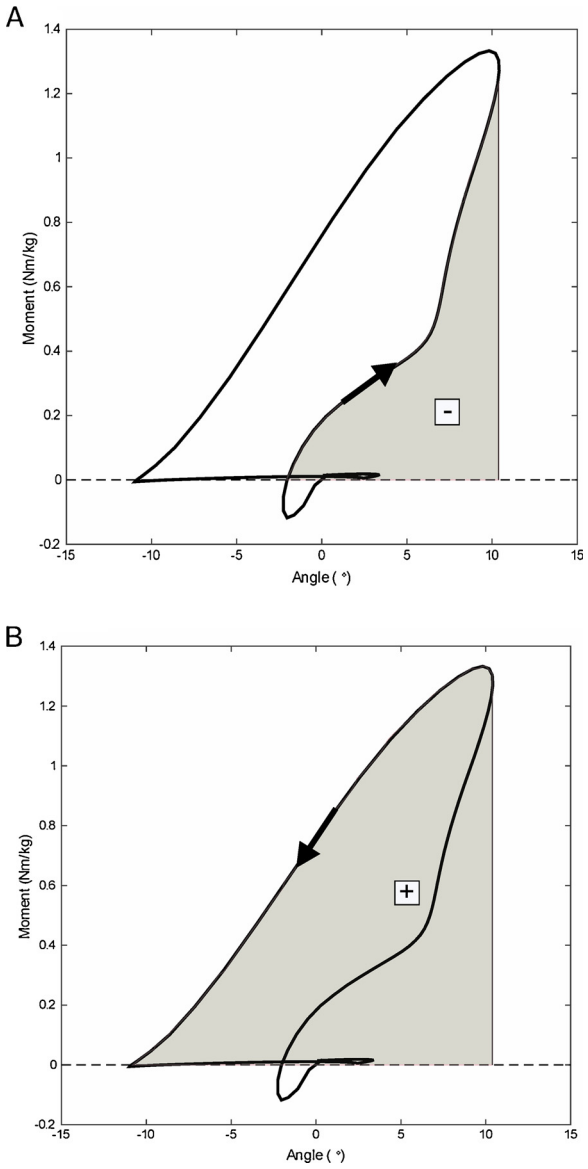

Fig. 2. Examples of moment-angle loops illustrating (A) work absorbed and (B) work produced (reference Crenna and Frigo 2011).

a long-term alteration in the gait pattern implying a reduced dorsiflexion moment in the ankle during early stance phase. A significantly lower ERP in ankle DJS has previously been reported in children with JIA (n = 8) when compared with healthy controls and children who were idiopathic toe walkers, but variations in walking disability were not assessed [15]. In that study, the LRP and DP did not differ between children with JIA and the control group.

The gait pattern exhibited by children with JIA shares several common characteristics with the gait pattern seen among adults with Rheumatoid Arthritis (RA), e.g. reduced walking speed and increased double support time [10], but at present, there is only one study on ankle DJS in RA. In that article [19], a different subdivision of the stance phase was used; 1) controlled plantar flexion (CPF), 2) controlled dorsiflexion (CDF), and 3) powered plantarflexion (PPF), making direct comparisons with the present material complex. However, the authors reported increased ankle DJS during CPF in women with RA, compared to age-matched healthy controls, and decreased ankle DJS during CDF in the most affected leg in the women with RA. An increased DJS has also been reported in patients with knee osteoarthritis (OA), where more advanced disease is associated with increased DJS in the knee during loading response irrespective of gait speed [20]. It has been shown that increased knee DJS persists six months after total knee arthroplasty, at a time when the postoperative pain has subsided [21]. It was suggested that the increased knee DJS was related to quadriceps weakness, or alternatively, a learned motor strategy that was adopted in the presence of weakness and pain but that persisted long after the pain and instability had resolved.

Thus, short-term and long-term alterations in DJS may be influenced by factors that do not relate primarily to the mechanical properties of the joint under study, e.g. neuromuscular adaptations to compensate for perceived pain, weakness or instability. Therefore, the term “quasi-stiffness” has also been proposed [22]. Patients with JIA and RA share several common gait adaptations. In a recent review on gait analysis of the lower limb in adults with RA [23] it is suggested that patients with RA walk slower in order to control the speed of heel strike and toe off and avoid pain. In the present study, when analyzing the unstratified data, there was a significant negative correlation between pretreatment ERP and pain while walking, at baseline and at three months, indicating a long-term adaptation of the gait pattern which leads to a reduced ankle dorsiflexion moment during early stance phase. Taken together, these findings imply that a decreased ERP represents a long-term alteration in gait pattern in children with JIA that may be associated with pain during walking.

One limitation of the present study is that group stratification is based on children’s responses to a questionnaire. Particularly for younger children, the capacity to understand questions and response

Table 1
Comparison of baseline demographics, disease parameters, and self-reported pain, health status, and disability between children with JIA reporting no walking difficulties (ND) and children with JIA reporting walking difficulties (WD).

	ND	WD	Significance
Number of subjects	13	20	
Gender, girls (%)	9 (69)	17 (85)	
Mean Age in years (SD)	10.6 (3.8)	12.0 (4.5)	n.s.
Mean Body weight in Kg (SD)	36.7 (14.3)	49.2 (24.0)	n.s.
Mean Body height in m (SD)	1.42 (0.18)	1.48 (0.25)	n.s.
Mean BMI (SD)	17.4 (2.8)	20.7 (4.9)	$p = 0.04$
Mean Disease duration in years, (SD)	3.3 (2.3)	5.6 (4.3)	n.s.
Median Joint Score (range)	5 (1-15)	10 (3-18)	n.s.
Median Pain (range) (VAS 0-100),	0 (0-60)	40 (0-79)	$p < 0.01$
Median CHAQ (range) (0-3 scale),	0.12 (0-0.75)	0.88 (0.38-2.12)	$p < 0.001$
Median CHAQ walking dimension (range) (0-2 scale)	0 (0)	1 (1-2)	$p < 0.001$
Median JAFI impairment (range) (0-36 scale)	3 (0-10)	18 (0-27)	$p < 0.001$
Median JAFI activity limitation (range) (0-56 scale)	3 (1-20)	27 (8-47)	$p < 0.001$
Median JAFI participation (range) (0-16 scale)	1 (0-5)	7 (0-16)	$p < 0.001$

[a] The significance level was set at $p < 0.05$. A higher p-value is referred to as not significant (n.s.).

Table 2
Comparisons of dynamic joint stiffness, work, and walking speed between children with JIA reporting no walking difficulties (ND) and children with walking difficulties (WD) before and three months after intraarticular corticosteroid joint injection (IACI) treatment.

	Pretreatment			3 months posttreatment		
	ND n = 13	WD n = 20	Significance	ND n = 13	WD n = 20	Significance
Mean Early Rising Phase, $Nm(kg*deg)^{-1}$, (SD)	0.05 (0.02)	0.03 (0.02)	$p < 0.05$	0.05 (0.03)	0.03 (0.01)	$p < 0.05$
Mean Late Rising Phase, $Nm(kg*deg)^{-1}$, (SD)	**0.24 (0.22)**	0.11 (0.06)	$p < 0.05$ [b]	0.09 (0.34)	0.10 (0.10)	n.s.
Mean Descending Phase, $Nm(kg*deg)^{-1}$, (SD)	0.08 (0.01)	0.07 (0.02)	n.s.	0.07 (0.01)	0.07 (0.02)	n.s.
Mean Work absorbed, $Nm(kg)^{-1}*deg$, (SD)	4.19 (2.72)	5.82 (2.48)	n.s.	5.77 (3.47)	6.01 (2.58)	n.s.
Mean Work produced, $Nm(kg)^{-1}*deg$, (SD)	10.01 (2.17)	9.96 (4.36)	n.s.	10.55 (2.77)	9.84 (4.02)	n.s.
Mean Work net, $Nm(kg)^{-1}*deg$, (SD)	5.10 (2.28)	4.13 (3.07)	n.s.	4.78 (2.24)	3.82 (3.23)	n.s.
Mean walking speed, $m*s^{-1}$, (SD)	1.19 (0.15)	1.15 (0.23)	n.s.	1.13 (0.12)	1.11 (0.21)	n.s.
Mean Non-dimensional walking speed (SD)	0.45 (0.07)	0.42 (0.08)	n.s.	0.43 (0.06)	0.41 (0.08)	n.s.

[a] The significance level was set at $p < 0.05$. A higher p-value is referred to as not significant (n.s.).
[b] Mann-Whitney.

options may be limited. Thus, we used a proxy version of the CHAQ for children below 10 years (completed by the same parent before treatment and after three months). The CHAQ is regarded as a valid and reliable tool for assessment of children with JIA [6]. In this study, some children with JIA reported no walking difficulties despite the fact they were receiving IACIs. It is important to note that IACIs are indicated when clinical signs of inflammation are present, regardless of walking difficulties as ankle arthritis in JIA is considered a predictor of disease progression. Another limitation if this study is the fact that the foot was modeled as a rigid segment and the ankle DJS analyzed only in the sagittal plane. Particularly in the later phases of stance, the ankle DJS will not mirror the mechanical behavior of other foot joints potentially affected by JIA. The relatively large standard deviation in LRP may be an indication of that. To investigate motion in other foot joints and in other planes, a multi-segmented foot model should be used.

4.1. Conclusion

Decreased ERP values in children with JIA and self-reported walking difficulties represent persistent changes in gait dynamics. Pretreatment gait analysis may aid in identifying children who will not have long-term benefit from IACIs in terms of improved gait, and therefore, may be informed and have the choice to be spared the risk of side effects associated with this treatment.

Declaration of interests

None.

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