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Knee joint kinematics of the pendulum test in children with and without Down syndrome

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Research Highlights:

- We studied the biomechanics of the pendulum test in children with and without DS.
- Children with DS had a lower first flexion excursion and a lower relaxation index.
- Children with DS had a lower mean and peak velocity and acceleration of leg swing.
- External ankle load reduced the medial-lateral knee shift in children with DS.
- Children with DS had less passive knee motion and increased joint stiffness.

Knee joint kinematics of the pendulum test in children with and without Down syndrome

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Abstract

Background: The Wartenberg pendulum test is a common clinical test that is used to measure stiffness about the knee in persons with and without disabilities such as cerebral palsy and Down syndrome (DS). Adults and adolescents with DS show fewer number of swing cycles and a lower relaxation index than healthy controls. However, it is not clear if children with DS show a similar trend compared to typically developing (TD) children.

Research question: Was the knee joint kinematics different between children with and without DS during the pendulum test?

Methods: Thirteen children with DS and 13 TD children participated in this study. There were two load conditions: no load (NL) and with ankle load (AL) equal to 2% of the subject's body weight. Five trials of a pendulum test were collected for each condition.

Results: The DS group showed a smaller first flexion excursion, a lower relaxation index, lower mean and peak velocities and accelerations during the first and second flexion and extension, and greater variability of acceleration during the first flexion than the TD group across both load conditions. This suggests that the DS group may have greater stiffness of the knee than the TD group to compensate for joint instability.

Significance: The pendulum test appears to be a valid test to evaluate the passive stiffness of the knee in children with DS. The lower relaxation index in children with DS suggests that larger bursts of quadriceps may be activated during a pendulum test, particularly in the first flexion excursion, to assure the knee joint stability.

Keywords: Leg swing; stiffness; ankle load; damping; medial-lateral shift.

1. Introduction

Nearly 1 in 800 infants are born with Down syndrome (DS) each year [1]. Individuals with DS exhibit significant delays in motor and cognitive development compared to their typically developing (TD) peers [2-4]. Impaired motor development in children with DS can result in reduced mobility due to ligament laxity [5, 6] and hypotonia [7, 8], which further reduces their social interaction and cognitive development. To overcome motor dysfunction, individuals with DS often develop compensatory strategies for locomotor tasks such as walking [9-14] and jumping [15] by increasing muscle activation or co-contraction [16-18] to increase stability of the joint [19].

The leg drop test, or better known as the Wartenberg pendulum test, was originally used to measure muscle tone in persons with Parkinson's disease [20]. This test has since been used to assess the passive stiffness of a joint in other populations such as stroke [21], cerebral palsy [22, 23], and DS [19, 24]. A joint is considered to have stiffness if reduced passive motion occurs at the joint, due to the intrinsic mechanical property of tissue to resist deformation [25]. Individuals with increased passive joint stiffness demonstrate decreased flexion during the first flexion swing, resulting in a lower relaxation index score, as well as lower peak and mean velocities. Previous studies showed that both adults [19] and adolescents [24] with DS show a lower relaxation index and the lower mean and peak velocities during the first flexion excursion than healthy controls. However, no study of the pendulum test has been conducted in children with DS.

Adding an additional load above the ankle has been shown to improve the kinetic pattern of vertical ground reaction force and facilitate leg swing and general muscular activation in children with DS during treadmill walking [11]. As additional ankle load increases the moment of inertia of the lower leg, this should increase the difficulty of the lower leg to resist the swinging motion and allow a greater amount of passive motion to occur, which may help children with DS elicit a leg swing pattern closer to that of TD children [26].

Therefore, the purpose of this study was to compare the knee joint kinematics between children with and without DS during the pendulum test with and without ankle load. We hypothesized that children with DS would demonstrate reduced knee passive motion than TD children. Children with DS would demonstrate a lower leg excursion and slower velocity during the leg swings. Additionally, the inclusion of an ankle load would reduce joint stiffness and improve the kinematics of leg swing.

2. Methods

2.1 Subjects

Fifteen TD children and 15 children with DS were recruited for this study. The inclusion criteria were that children were between the ages of 6 and 12 with a diagnosis of DS. The exclusion criteria were that they had uncorrected cardiovascular diseases, uncorrected visual or hearing problems, an injury to their legs or any seizures within past six months, or any other medical problems that prevented them from participating in this study. Due to technical problems, data from only 13 TD children and 13 children with DS are available for data analysis. The DS group had a shorter height, a shorter lower leg length, and a lower body mass than the TD group (Table 1). The hosting university institutional review board approved all test procedures. Prior to data collection all subjects and parents were informed about the procedures and aims of the study. Parents signed a parental permission form and children provided verbal assent.

2.2 Procedures

For each subject, we collected anthropometric data including leg dominance, body mass, height, leg length, and lower leg length. Leg dominance was determined by having each subject kick a soccer ball on the floor. The leg that was used to kick the ball was established as the dominant leg. Leg length was measured from the anterior superior iliac spine to the medial malleolus of the tibia while standing upright. Lower leg length was measured from the medial condyle of the tibia to the floor. An eight-camera Vicon motion capture system (Vicon, Oxford, UK) was used to record 3D movement of the leg at the frequency of 100 Hz. Sagittal-plane knee angles and frontal-plane movements of the leg were calculated using reflective markers placed on the lateral side of the thigh, knee, and tibia. The thigh marker was placed on the 1/3 proximal length of the femur. The knee marker was placed on the flexion-extension axis, which was determined by having each subject perform slow squats. The tibia marker was placed on the 1/3 proximal length of the lower leg.

Subjects were seated upright at the edge of a massage table with their legs hanging over the edge of the table [20] . Each subject's dominant leg was lifted by the examiner, so that the knee was in full extension (Fig. 1a, point 1). They were instructed to relax their leg, and allow it to swing freely without voluntary control until it stopped swinging [20] (Fig. 1a, point 2). There were two load conditions: no load *(NL)*, and ankle load *(AL*) equal to 2% of their body mass. The ankle load increases the moment of inertia of the lower leg about the knee by 60.7% [27] and was the outer limits for children with DS [11]. We attached an adjustable weight cuff above the ankle and strapped it tightly to minimize any motion during leg swing. Each subject completed five successful trials for each condition with five seconds between trials. Trials were considered successful when the subject allowed their leg to swing freely, did not actively swing their leg, and did not produce additional movements such as raising their thigh or hips from the table.

2.3 Measurements

The 3D data from each trial were first rotated with respect to the orientation of the lower leg so that the coordinates of the new data represented the medial-lateral (ML), anterior-posterior (AP), and vertical directions, respectively. The data were filtered using a 4th order zero-lag low-pass Butterworth with a cut-off frequency of 6Hz [28]. Kinematic variables were then calculated (Fig. 1b) including: the first flexion excursion (A_1), relaxation index (RI), and number of swing cycles. RI was calculated as $RI=A_1/A_0$, where A_0 is the difference between the starting angle and the resting angle [19]. The number of swing cycles were counted as the number of times between two peaks of flexion until the lower leg reached a resting state [25] with a cutoff of 3° towards extension [23]. Natural frequency (ω) was calculated as a ratio of stiffness to mass according to Fee and Miller [22],

$$\omega = 2\pi/T$$

where *T* is the period of one cycle. A damping ratio (ζ), damping coefficient (B), and stiffness coefficient (K) were calculated as below [19] between the first and second angles of reversal at the end of flexion (F1 and F2):

$$\zeta = \sqrt{\frac{(lnD)^2}{4\pi^2 + (lnD)^2}}$$

where *D* is the ratio of the peak angle of one cycle to the peak angle of the next cycle.

$$B = 2 \cdot \zeta \cdot \omega \cdot J$$

where *J* is the moment of inertia about the knee axis of rotation.

$$K = J \cdot \omega^2$$

J and mass characteristics (lower-leg-foot segment) were calculated for each subject according to Winter [27].

We graphed a phase portrait (angular velocity versus angular displacement) to explore differences in the dynamics of leg swings between the two groups [23, 29, 30]. The “whirlpool” shape gives an indication of joint relaxation (Fig. 1 e & f). For healthy subjects, the shape of the whirlpool is uniform. Any voluntary input from the subject results in a disruption in the uniform shape of the whirlpool [30]. Additionally, we plotted the acceleration of the first flexion excursion over time to understand possible synchronous bursts of muscle activity [24].

To quantify any motion in the front plane, medial and lateral shift of the knee were calculated. The medial and lateral shift was calculated by using the initial position of the knee marker, then subtracting it from the minimum and maximum value in the horizontal direction, respectively. Additionally, to account for possible contribution from the hip, an angle formed by the thigh, knee and tibia markers projected to the transverse plane was calculated. When the motion of the swinging leg occurs purely in the sagittal plane, the knee and tibia markers are in line. When the leg is internally rotated, the knee and tibia are medially and laterally displaced, respectively. Therefore, the angle formed between the line formed by the knee and tibia markers and the line formed by the knee and thigh markers on the anterior-posterior axis was calculated as a projection of internal and external rotation of the hip. This angle does not represent true internal and external rotation about the hip, but to measure the contribution from the hip to the pendulum test. All calculations were performed using MATLAB version R2015a customized programs (Mathworks, Natick, MA).

2.4 Statistical Analysis

A series of two-way (2 group x 2 load) mixed ANOVAs with repeated measures on load were performed for the first flexion excursion, RI, peak and mean velocities, peak accelerations, damping ratio, natural frequency, stiffness coefficient, and damping coefficient. Post-hoc pair-wise comparisons were conducted when necessary. Normality was checked for each variable. When appropriate, log transformations were conducted for the variables that were not normally distributed. Statistical analyses were performed using SAS, version 9.4 (SAS Institute Inc., Cary, NC). A significance level was set at $\alpha=0.05$ for all tests.

3. Results

The DS group displayed a smaller number of cycles, a smaller first flexion excursion, and a lower RI than the TD group (Fig. 1c and d, Table 2). Specifically, the DS group had an average first flexion excursion of 54.02° and an average RI of 1.33 compared to an excursion of 74.25° and a RI of 1.74 in the TD group across both load conditions. For the phase portraits, the TD showed a smooth "whirlpool" display with no additional "whirls" in the center nor clustered whirls (Fig. 1e). The DS group, however, demonstrated some clustering of the "whirls" (Fig. 1f). Statistical analysis revealed that there was a group effect on the first flexion excursion ($p<0.001$, see Table 2 for details) and RI ($p=0.006$). In addition, there was a trend for a load effect on the stiffness coefficient in favor of ankle load ($p=0.063$) across the two groups.

In the frontal plane, there was a load effect on the lateral shift of the knee ($p=0.036$, Table 2) and a group by load interaction on the ROM of the knee shift ($p=0.033$). Specifically, the DS group reduced the ROM of the knee shift from no load to ankle load while the TD group maintained their values.

Across both load conditions, the DS group displayed a slower peak velocity than the TD group for the first and second flexion and extension of the leg swing and a slower mean velocity than the TD group for the first flexion and extension (Fig. 2). Statistical analysis demonstrated that there was a group effect in favor of the TD group in peak velocity of the first flexion ($F(1,24)=21.80$, $p<0.001$) and the second flexion ($F(1,24)=28.55$, $p<0.001$, Fig. 2a), of the first extension ($F(1,24)=21.97$, $p<0.001$) and the second extension $F(1,23)=22.39$, $p<0.001$, Fig. 2b), as well as in mean velocity of the first flexion ($F(1,24)=26.31$, $p<0.001$) and the first extension ($F(1,24)=17.66$, $p<0.001$, Fig. 2c). Additionally, there was a load effect on peak velocity for the second flexion ($F(1,21)=7.09$, $p=0.015$) and second extension ($F(1,21)=18.01$, $p<0.001$) such that both groups increased their peak velocities from no load to ankle load condition.

For the acceleration of the swinging leg, the DS group showed a similar trajectory but greater variability than the TD group (Fig. 3). Both groups demonstrated an acceleration of the limb until around 40% of the first flexion excursion and decelerate thereafter. The two groups differed during the 70% to 90% of the first flexion excursion such that the TD group demonstrated a greater deceleration of the lower leg while the DS group did not. Specifically, the DS group displayed a lower second peak acceleration ($F(1,24)=16.64$, $p<0.001$, Fig. 4) than the TD group.

4. Discussion

Our study revealed that children with DS have increased passive stiffness of the knee compared to the TD group. This is consistent with previous findings of kinematic data in adults and adolescents with DS [19, 24]. The clustering of “whirls” in the phase portrait for the DS group (Fig. 1f) suggests that there might be activation of the quadriceps during the pendulum test. Additionally, a less acceleration of the leg into the first flexion in the DS group (Fig. 3) suggests that there may be activation of the quadriceps. Further, our data show that there is possibly medial-lateral shift of the knee during the pendulum test and additional contribution from the hip that could be contributing to the reduced passive motion about the knee in the children with DS.

4.1 Passive stiffness and sagittal plane motion

Consist with our hypothesis, the DS group showed increased knee joint stiffness evidenced by decreased first flexion excursion and RI compared to the TD group. Similar results were also reported in adults with DS [19] and adolescents with DS [24], where higher levels of quadriceps activation were observed during the first flexion excursion that corresponded with lower accelerations of the leg. It was concluded that individuals with DS might have actively contracted their quadriceps to stabilize the knee joint [5, 6].

Our results suggest that individuals with DS may be at a higher risk of limited motor function from an early age. Reductions in mobility can restrict children with DS from participating in physical activity, which causes motor abilities to diminish as they age [2, 4]. Typically, RI scores equal to or greater than 1.6 indicate that there is no hyperactive muscle activity. The TD group did not show indications of stiffness whereas the DS group did during both load conditions.

The inclusion of the ankle load during the pendulum test showed promising results. Our results supported our hypothesis such that the inclusion of the ankle load improved the joint stiffness in both groups by increasing the number of cycles and stiffness coefficient, particularly the DS group. While the DS group showed lower RI and A1 values than the TD group, the stiffness and damping coefficients were not different between the groups. Similar findings were reported in stiffness coefficients between adults with and without DS [19] and damping coefficients between adolescents and adults with DS [24]. Additionally, the natural frequency and damping ratio were not significantly different between our two groups (Table 2). The lack of a statistical difference for the stiffness and damping coefficients between the two groups may be a result of inter-subject variability [19].

4.2 Leg swing motions in other planes

Our results demonstrate that there is a relatively large amount of movement of the lower leg in the frontal plane. This was supported by the projection angle formed by the knee and tibia and the amount of knee shift in the medial and lateral direction. A trend for group significance in the amount of projected internal thigh rotation (p=0.060) suggests that some hip muscles might possibly contribute to the knee kinematics during the Wartenberg pendulum test. The inclusion of the ankle load decreased the ROM shift of the knee in the DS group, suggesting ankle load might help reduce the contribution of the hip muscles during the pendulum test.

The movement in the frontal plane can also be attributed to the mass distribution of the thigh. As the mass of the thigh is not distributed evenly from the hip to the knee joint, it increases the likelihood of circumduction about the hip once the leg is released. While both the upright and supine positions are commonly used, they may limit the passive motion of the lower leg due to the anatomical length of the hip flexors and quadriceps at these two positions. Having the hip in a neutral position such as an inclined position may provide the more reliable results. Further study is warranted to examine the effect of body positions on the pendulum test.

4.3 Limitations

First, we were not able to collect EMG data for most of our subjects due to their skin sensitivity. This limited our ability to determine the neuromuscular cause of the stiffness during the leg swings. It also prevents us from comparing our child subjects to adults and adolescents with DS. Second, due to the delays in cognitive development in children with DS, our results may be biased in favor of the TD group. Third, the use of ankle load during this test did provide some beneficial information. However, since there was only one ankle load condition, this limited our ability to determine an optimal load for children with DS. Nonetheless, ankle load equal to 2% of body mass induced some changes in the knee stiffness. Additional investigation into larger external loads is warranted to determine if this trend continues with larger external loads and if this task can ultimately be used as an intervention tool.

5. Conclusion

Children with DS demonstrate greater joint stiffness during the pendulum test, likely due to their increased ligament laxity and unnecessary muscle activation. Therefore, it is important to focus therapy interventions that address the issue of reduced passive motion. Strengthening the muscles around the joint may increase joint stability; however, that alone may not resolve this issue. The inclusion of the external load during the pendulum test showed promising results and may facilitate the performance of the pendulum test in children with DS.

Conflict of Interest

None.

Funding

None.

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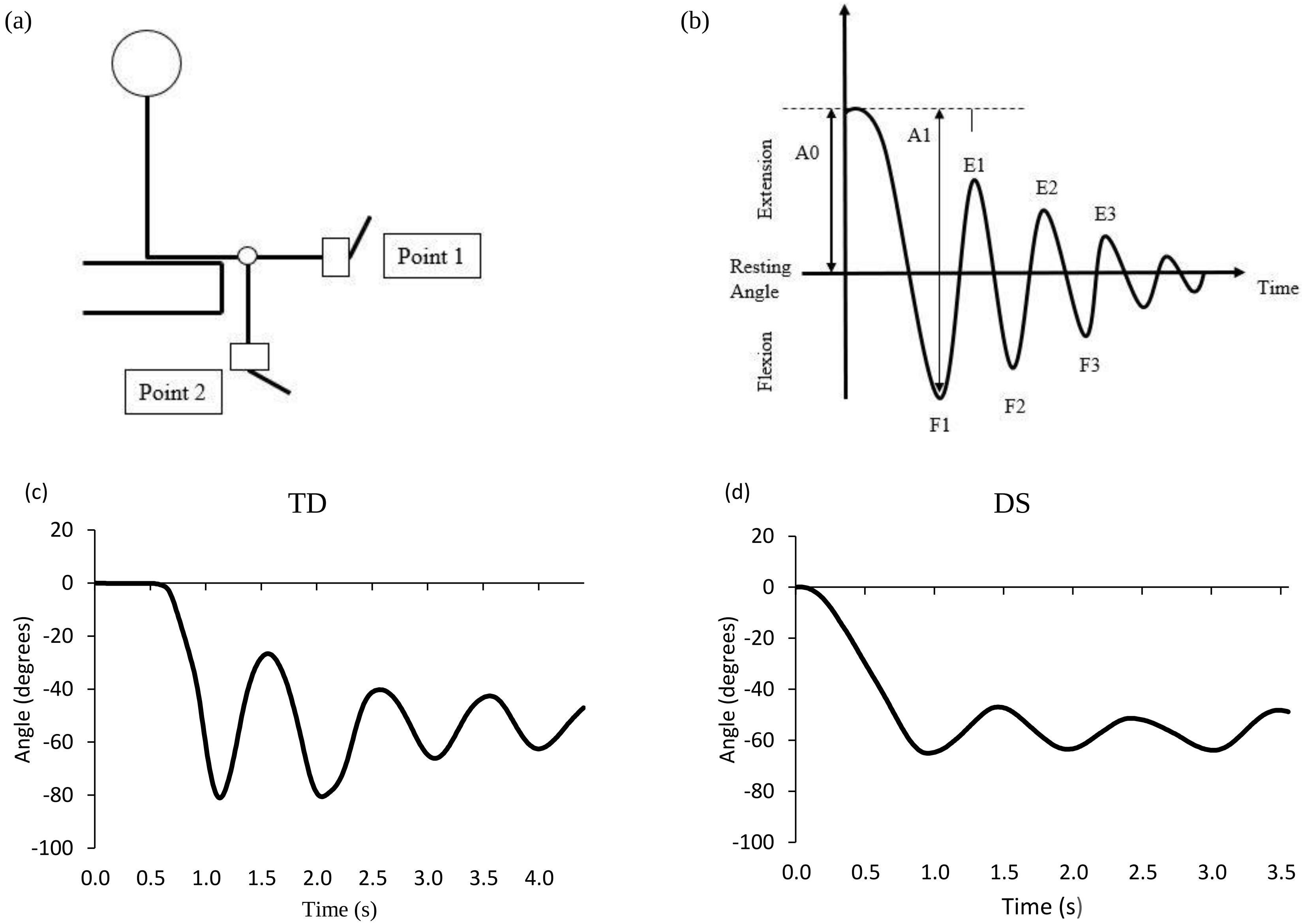
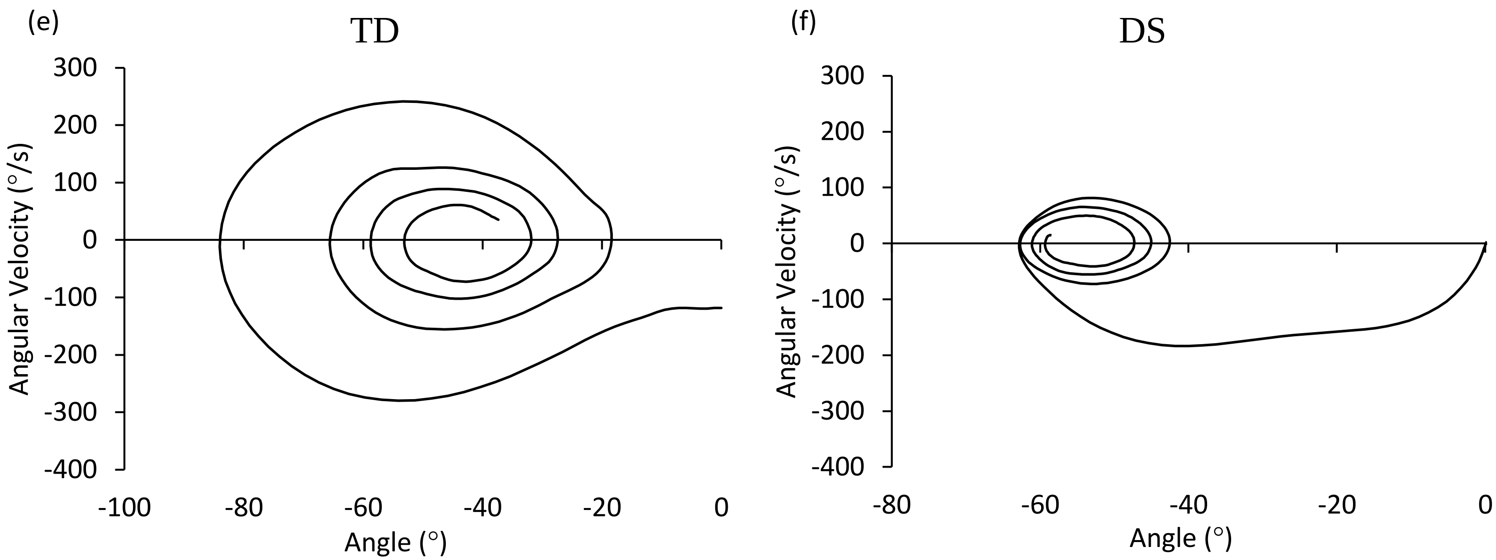
Figures

Fig. 1:





(e)
TD
300
200
100
0
-100
-200
-300
-400
Angular Velocity (°/s)
-100
-80
-60
-40
-20
0
Angle (°)
(f)
DS
300
200
100
0
-100
-200
-300
-400
Angular Velocity (°/s)
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-60
-40
-20
0
Angle (°)

Fig. 2:

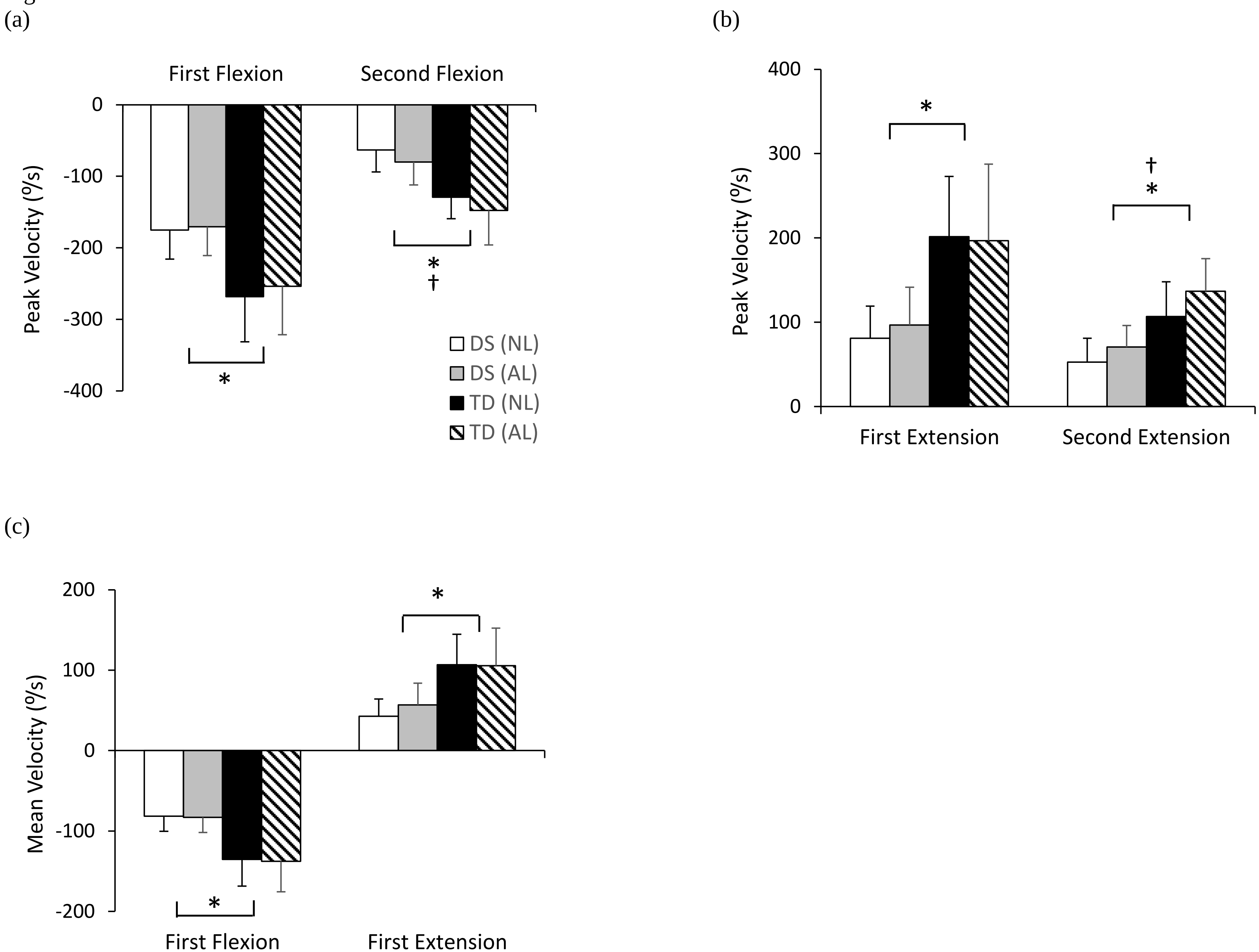

(a)
First Flexion
Second Flexion
Peak Velocity (º/s)
0
-100
-200
-300
-400
*
*
†
DS (NL)
DS (AL)
TD (NL)
TD (AL)
(b)
Peak Velocity (º/s)
400
300
200
100
0
*
†
*
First Extension
Second Extension
(c)
Mean Velocity (º/s)
200
100
0
-100
-200
*
*
First Flexion
First Extension

Fig. 3:

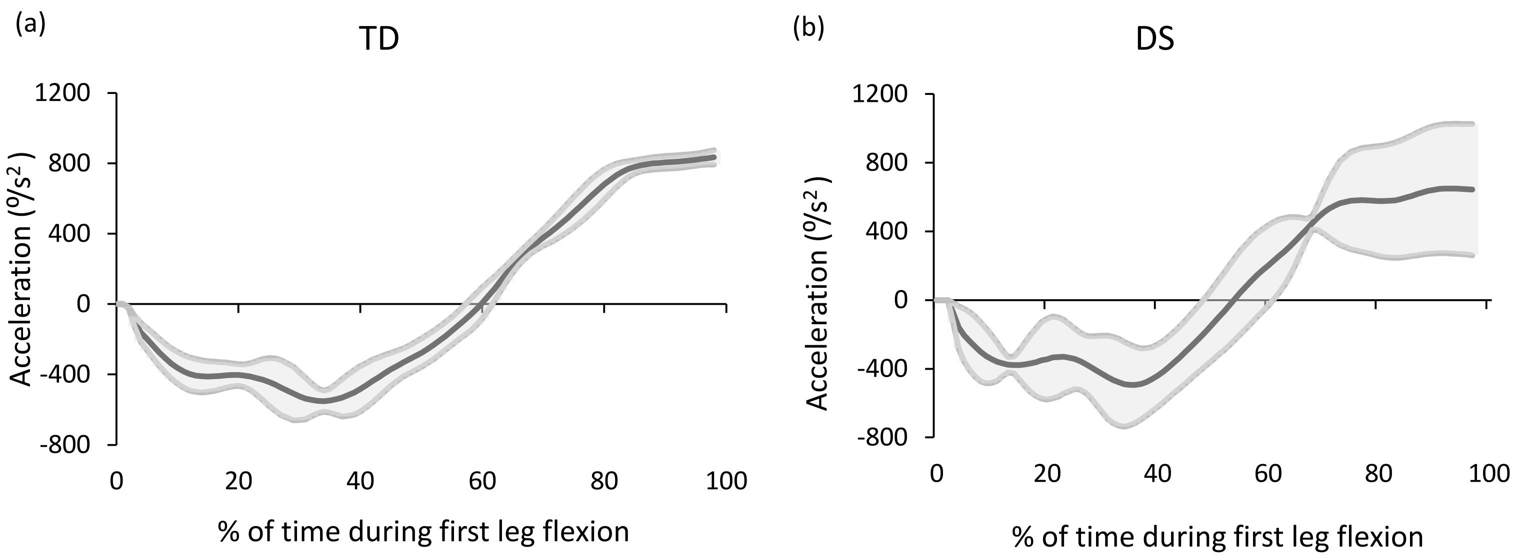
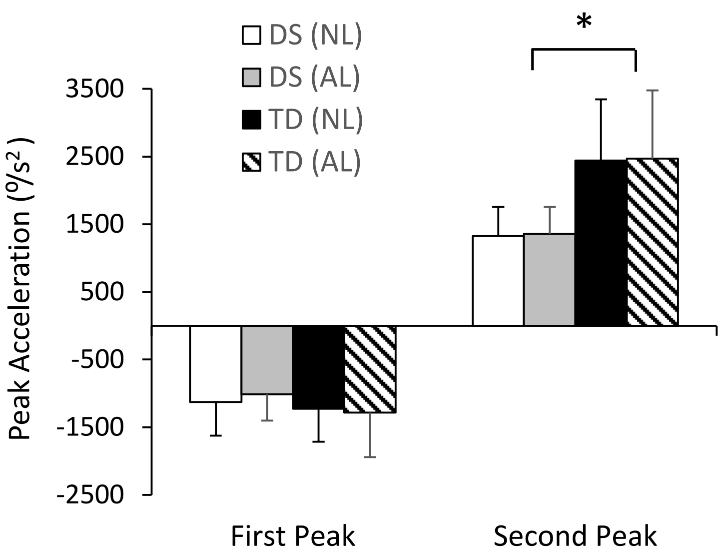

(a)
TD
1200
800
400
0
-400
-800
Acceleration (⁰/s²)
0
20
40
60
80
100
% of time during first leg flexion
(b)
DS
1200
800
400
0
-400
-800
Acceleration (⁰/s²)
0
20
40
60
80
100
% of time during first leg flexion

Fig. 4:


DS (NL)
DS (AL)
TD (NL)
TD (AL)
*
3500
2500
1500
500
-500
-1500
-2500
Peak Acceleration (⁰/s²)
First Peak
Second Peak

Figures Captions

Figure 1: The Wartenburg pendulum test. (a) Visual demonstration of the test. A subject sits at the edge of a table with his legs hanging over the edge of the table. The examiner raises the leg being tested to the full knee extension position (Point 1). The subject relaxes his leg, and the examiner releases the leg. The subject allows his leg to swing freely until it comes to a stop (Point 2). Ankle load placement is represented by the rectangle. (b) Graphic representation of the typical pattern of the pendulum test. F1 and F2 are the angles of reversal at the end of the first and second flexion, respectively. E1 and E2 are the angles of reversal at the end of the first and second extension, respectively. A_0: the starting knee extension angle with respect to the resting angle; A_1: the first swing excursion from the starting knee extension to the maximal knee flexion (F1). Relaxation index (RI) is calculated as the ratio of A_1/A_0. Number of swings was counted as the number of times between two peaks of flexion until the lower leg reached a resting state, with a cutoff of 3° towards extension. Representative plots of the knee joint kinematics in the sagittal plane under no load for (c) a typically developing (TD) subject and (d) a subject with Down syndrome (DS). Phase plots of angular velocity against angular displacement for (e) the TD subject and (f) the DS subject. These “whirlpool” plots give an indication of any voluntary input from the subjects. A uniform pattern is associated with healthy subjects and voluntary input is detected by alterations in the pattern.

Figure 2: Mean and standard deviation of velocity profiles in the sagittal plane. (a) Peak velocities of the first and second flexion, (b) peak velocities of the first and second extension, and (c) mean velocities for the first flexion and first extension. Negative values represent

flexion and positive values represent extension. DS: Down syndrome, TD: typically developing. AL: Ankle load, NL: No Load. A symbol * denotes a group effect at $p<0.05$. A symbol † denotes a load effect at $p<0.05$.

Figure 3: Representative plots of acceleration profile for (a) one TD subject and (b) one subject with DS during the first flexion excursion. The mean values (black lines) and ± 1 standard deviation (grey lines) were calculated for the “No Load” condition for each subject. DS: Down syndrome, TD: typically developing.

Figure 4: Mean and standard deviations of peak acceleration profiles in the sagittal plane. DS: Down syndrome, TD: typically developing. AL: Ankle load, NL: No Load. A symbol * denotes a group effect at $p<0.05$.

Table

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Conflicts of Interest Statement

There were no conflicts of interest when completing this study.

Author contribution

Diego Ferreira contributed to Data curation; Formal analysis; Investigation; Methodology; Software; Validation; Visualization; Original draft writing; Review and editing.

Huaqing Liang contributed to Data curation; Formal analysis; Original draft writing; Review and editing.

Jianhua Wu contributed to Conceptualization; Data curation; Formal analysis; Funding acquisition; Investigation; Methodology; Project administration; Resources; Software; Supervision; Validation; Visualization; Original draft writing; Review and editing.