Quantifying lower limb inter-joint coordination and coordination variability after four-month wearing arch support foot orthoses in children with flexible flat feet

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ABSTRACT

Introduction: Flat feet in children negatively affect lower limb alignment and cause adverse health-related problems. The long-term application of foot orthoses (FOs) may have the potential to improve lower limb coordination and its variability.

Aim: To evaluate the effects of long-term use of arch support FOs on inter-joint coordination and coordination variability in children with flexible flat feet.

Methods: Thirty boys with flexible flat feet were randomly assigned to the experimental (EG) and control groups. The EG used medial arch support FOs during daily activities over a four-month period while the control group received a flat 2-mm-thick insole for the same time period. Lower-limb coordination and variability during the 3 sub-stance phases were quantified using a vector coding technique.

Results: Frontal plane ankle-hip coordination in EG during mid-stance changed to an anti-phase pattern (156.9°) in the post-test compared to an in-phase (221.1°) in the pre-test of EG and post-test of CG. Frontal plane knee-hip coordination in EG during LR changed to an anti-phase pattern (116°) in the post-test compared to an in-phase (35.5°) in the pre-test of EG and post-test of CG. Ankle inversion/eversion-knee internal/external rotation joint coupling angle in EG changed to an in-phase pattern (59°) in the post-test compared to a proximal phase (89°) in the pre-test. Coupling angle variability increased in the post-test of EG for sagittal plane ankle-hind during push-off, transverse plane ankle-hind during LR and mid-stance, and transverse plane knee-hind during LR and mid-stance compared to pre-test of EG and post-test of CG.

Conclusion: The long-term use of arch support FOs proved to be effective to alter lower limb coordination and coordination variability during walking in children with flexible flat feet. This new insight into coordinative function may be useful for improving corrective exercise strategies planned for children with flat feet.
1. Introduction

Flat foot is reported in 4% of ten-year-old children and 10% of these individuals showed flexible flat feet requiring treatment to prevent secondary deformities in their adulthood (Kosashvili, Fridman, Backstein, Safrir, & Ziv, 2008; Riskowski et al., 2013). Flexible flat foot is a very common condition in children (Rome, Ashford, & Evans, 2010) in which the medial longitudinal arch of the foot largely collapses during the stance phase of gait and restores after removal of body weight (Pourhoseingholi & Pourhoseingholi, 2013). Rigid flatfoot, on the other hand, is characterized by a stiff and flattened foot arch during both weight bearing and non-weight bearing conditions (Iodice, Bellomo, Migliorini, Megna, & Saggini, 2012). Although treatment of the flexible flat feet by non-surgical methods is relatively efficient, the treatment of the rigid flat feet is difficult and needs surgical interventions (Vukasinovic et al., 2011). Children with flexible flat feet are prone to having pain or trouble at the knee, hip and back (Kothari, Dixon, Stebbins, Zavatsky, & Theologis, 2016). Various types of interventions like foot orthoses (FOs) are used to treat these problems and are commonly utilized in the clinical setting (Vicenzino et al., 2008; A. E. Williams, Hill, & Nester, 2013).

Arch support FOs are made from a negative cast based on the subtalar joint neutral position (Michaud, 1993). Accordingly, the position in the cast might enable the subtalar joint to be in a neutral position immediately after heel strike and at the midstance phase of gait. It is believed that wearing FOs may realign the lower-extremity (Telfer, Abbott, Steultjens, & Woodburn, 2013) and also may affect lower extremity joint coordination (Eslami & Ferber, 2013).

Recent studies focused on high-order variables such as inter-joint coordination and coordination variability, as it is believed that coordination between joints or segments may distinguish motor behaviors. Moreover, coordination between adjacent segments has been implicated in the development of injuries such as iliotibial band syndrome (Hamill, Van Emmerik, Heiderscheit, & Li, 1999; Miller, Meardon, Derrick, & Gillette, 2008). Evaluating inter-segment coordination allows us to perceive how motor systems collaborate to achieve a movement task. This evaluation provides additional information to the joint kinematics. The information derived from joint coordination variability could be also useful to determine movement flexibility and stability. Previous studies suggest that coordination variability could have a functional role in controlling strategies applied to a motor system and enhance adaptability to motion perturbations and task limitations (Davids, Glazier, Araujo, & Bartlett, 2003; Seifert, Button, & Davids, 2013).

Continuous relative phase (CRP) and vector coding are two common methods for assessing joint coordination and coordination variability. The use of CRP limits the analysis of coordination to the phase relationship between two segments. On the other hand, vector coding and four coordination phases indicating the relationship between two movements as well as in-phase or anti-phase of movements (Chang, Van Emmerik, & Hamill, 2008) provide valuable information about motor control and dominancy of one movement or segment over another. This information can be of assistance in a clinical setting (Seay, Van Emmerik, & Hamill, 2011).

Previous studies reported different kinematic coupling behavior between individuals with pes planus and pes cavus foot (Nawoczenski, Saltzman, & Cook, 1998) as well as between individuals with excessive and normal subtalar pronation (McCay & Manal, 1997). Accordingly, few studies have investigated the immediate effects of FOs on lower limb joint coordination. A previous study demonstrated that healthy females have a more coordinated relationship (in-phase) and greater coordination variability of foot/shank during landing with FOs than that landing without FOs (Noghondar & Yazdi, 2017). FO stiffness, however, has no significant effect on the coordination pattern and coordination variability of foot-shank and shank-thigh during landing (Noghondar & Yazdi, 2017). On the other hand, wearing FOs can play a major role in the maintenance of coordination variability of the tibia (transverse plane) and calcaneus (frontal plane) coupling during running (MacLean, Van Emmerik, & Hamill, 2010). Hamill, Bates, and Holt (1992) argued that running in shoes with a soft midsole construction leads to a decoupling of knee flexion and rearfoot eversion and a series of antagonistic counter-rotations. Furthermore, it was demonstrated that wearing varus posted running shoes could alter foot–shank coupling towards a distal-phase coordination pattern (Van Woensel & Cavanagh, 1992). Nawoczenski, Cook, and Saltzman (1995) examined the effect of FOs on the coupling coefficient in individuals with high and low arched feet. The ratio significantly increased when the custom FOs were worn by subjects in both groups. Eslami and Ferber (2013) examined 3D kinematic changes in forefoot-rearfoot coupling for adults with different navicular drop measures during running and reported that wearing semi-rigid FOs significantly decrease the forefoot-rearfoot joint coupling angle by reducing forefoot frontal plane motion relative to the rearfoot motion. According to the abovementioned studies, kinematic coupling is different between normal foot and flat foot and likewise, FOs may alter gait biomechanics in flat feet patients to exhibit the same inter-segmental coordination as healthy individuals.

In general, FOs are clinically used for a long-term period and it can take a certain period of time for patients to get comfortable with wearing them (Moisan & Cantin, 2016). However, most studies only quantify instantaneous effects of wearing FOs on biomechanical variables or their effects after a short-term period (2 weeks or less) (Dedieu, Drigeard, Gijini, Dal Maso, & Zanone, 2013; Mündermann, Wakeling, Nigg, Humble, & Stefanyszyn, 2006; Murley, Landorf, & Menz, 2010; Tomaro & Burdett, 1993), and have not quantified whether inter-joint coupling angles and their coordination variability change over time. It is therefore unknown whether wearing arch support FOs in children with flexible flat feet for a certain period of time induces inter-joint coordination and their coordination variability adaptations.

Few studies have provided evidence whether an orthotic device can affect gait mechanics in children with flexible flatfeet (J. J. Eng & Pierrynowski, 1994; Jafarneshadgero, Madadi Shad, & Ferber, 2018; Kothari et al., 2016; Pauk & Ezerskiy, 2011; Twomey & McIntosh, 2012). For example, Jafarneshadgero et al. (2018) reported that FOs decrease frontal plane hip joint moment asymmetry during gait in children with flexible flatfeet. Twomey and McIntosh (2012) reported that children with flat feet showed increased hip external rotation coupled with increased external foot progression angle during walking, as compared to healthy controls. Kothari et al. (2016) stated that children with flat feet aged between 8 and 15 years old express pain or discomfort at the knee, hip and back during gait. In this condition, finding a solution to decrease the level of pain and discomfort is vital in both short-term and long-term patient care. If specific footwear interventions can modify gait biomechanics and inter-joint coordination in children with flat feet,
these may be helpful towards the management of symptoms in children with flat feet, thus reducing the risk of future injuries. No study to date has quantified the effects of long term use of arch support FOs on inter-joint coordination and their coordination variability in children with flexible flat feet. The principal objective of this study was therefore to evaluate the effects of wearing arch support FOs for four months on the lower extremity inter-joint coordination and coordination variability in children with flexible flat feet during walking. We hypothesized that long-term use of arch support FOs would modify the lower limb inter-joint coordination towards in-phase patterns (especially the ankle inversion/eversion and knee internal/external rotation joint coupling angle) (Noghondar & Yazdi, 2017). We also hypothesized that FOs would not affect lower limb inter-joint coordination variability, which is consistent with a previous research suggesting that insoles have no impact on inter-joint coordination variability of the lower extremity (Noghondar & Yazdi, 2017).

2. Methods

2.1. Participants

An a priori power analysis (G-power) revealed that for a statistical power of 0.80 and an effect size of 0.80 with an alpha level of 0.05, a sample size of at least 30 subjects was required (Faul, Erdfelder, Lang, & Buchner, 2007). Therefore, thirty male children with flexible flat feet were recruited for this study. All participants and their parents gave written permission to participate in this study. Navicular drop, resting calcaneal stance position and arch height index (AHI) were all measured to determine flexible flat feet. AHI is defined as the ratio of dorsal height at 50% of total foot length, divided by the foot length from the back of the heel to the head of the first metatarsal, defined as the truncated foot length. Individuals whose both feet had > 10 mm of navicular drop (Cote, Brunet, Gansneder, & Shultz, 2005), > 4° of eversion in calcaneal stance position (Bok, Kim, Lim, & Ahn, 2014) and with an arch height index less than 0.31 (Butler, Hillstrom, Song, Richards, & Davis, 2008) were admitted as subjects of the study. A subject was excluded from the study if he had any previous history of bone fractures, surgery, orthopedic disease, neuromuscular problems, limb length discrepancies of greater than 5 mm, or a self-reported feeling of fatigue, or having had heavy physical tasks or exercises during the past two days. The subjects were all right foot dominant determined through a kicking ball test. In this study, the subjects were randomly

![Flow diagram of the randomized controlled trial.](image-url)
allocated to two equal-sized groups (experimental group (EG) and control group (CG)) (Fig. 1). During the randomization procedure, a set of sealed, opaque envelopes, was used to ensure the concealment of the allocation. Each envelope contained a card stipulating to which group the child would be allocated. Neither the children nor their parents were aware of the group to which the subjects were allocated, thereby characterizing a blind study for the placebo effect of the insole in the CG. The CG (age: 10.4 ± 1.5 years; height: 141.2 ± 6.1 cm; mass: 48.2 ± 5.4 kg; body mass index: 20.1 ± 4.2 kg/m²; Navicular drop: 13.1 ± 1.9 mm; AH1: 0.18 ± 0.06; calcaneal eversion: 7.1 ± 0.9°) wore insoles without corrective elements and the EG (age: 10.5 ± 1.4 years; height: 142.4 ± 5.7 cm; mass: 48.1 ± 9.1 kg; body mass index: 20.0 ± 4.0 kg/m²; Navicular drop: 13.0 ± 2.1 mm; AH1: 0.18 ± 0.07; calcaneal eversion: 7.2 ± 1.1°) wore insoles with corrective elements (arch support FOs). Ethics approval was obtained from the Research Ethics Board of the University (ARUMS-REC-1396-90).

2.2. Apparatus

Kinematic data were collected at 100 Hz using a Vicon MX Motion Systems consisting of six T-series cameras (Vicon Motion Systems, Oxford, UK) and 16 spherical reflective markers with a diameter of 15 mm. The size of the cubic calibration volume was 4.0 m (length) × 2.0 m (width) × 2.0 m (height) that was located at the middle of a 15 m walkway. The Plug-in-Gait marker set was used to identify the bilateral pelvis, thighs, legs, and feet provided in Nexus software. Two force plates (Kistler, type 9281, Kistler Instruent AG, Winterthur, Switzerland) were used to record the kinetic data under each leg at 1000 Hz and were synchronized with the Vicon system. The force plates were located at the center of the calibrated volume.

2.3. Gait analysis

Before the initiation of experimental procedures, anthropometrical parameters of each subject were measured from selected anatomic landmarks for entering into Nexus software. Then, reflective markers were placed bilaterally to subjects as follows: anterior superior iliac spine, posterior superior iliac spine, lateral mid-thigh, lateral femoral epicondyle, mid shank, lateral malleoli, heel and superior iliac spine, posterior superior iliac spine, lateral mid-thigh, lateral femoral epicondyle, mid shank, lateral malleoli, heel and between second and third metatarsal heads. Then, subjects were asked to walk a number (three) of trials along the test walkway to familiarize themselves with the experimental surroundings. In addition, prior to each experiment condition, a static trial was captured to identify joint center locations and calculate the segment coordinate systems. During pre-test, data were collected during walking with shoes without inserting any foot orthoses into it while during post-test data were collected during walking with shoes with inserting foot orthoses into it. During pre and post-test, six walking trials were performed at self-selected walking velocity. A trial was accepted when subjects walked with no visible alteration in gait mechanics; whereas, it was excluded in case the foot was placed on the edge of the force plate.

GRFs data was filtered using a fourth-order low-pass Butterworth filter with a 20 Hz cutoff frequency. Kinematic data were filtered by zero-lag fourth-order low-pass Butterworth filter with a cut off frequency of 6 Hz. Kinematic data were calculated during the stance phase of walking which was defined as the interval from ground contact (vertical GRF > 10 N) to toe off (vertical GRF < 10 N) (Madadi-Shad, Jafarnezhadgero, Zago, & Granacher, 2019). Kinematic and GRF data were synchronized using Nexus software (Oxford Metrics, Oxford, UK). The gait cycle was divided into the following phases: loading phase (0–20% of gait cycle), mid-stance (20–47% of gait cycle), push off (47–70% of gait cycle), and swing phase (70–100% of gait cycle) (Jafarnezhadgero, Fatollahi, Amizadeh, Siahkouhian, & Granacher, 2019). Data for each stride was time-normalized to 100 data points using linear interpolation technique (David A. Winter, 2009). Graphical reports were created in Polygon Authoring Tool (PAT). Data were exported from PAT to a spreadsheet for patterns, ranges of motion and other specific data point calculations.

2.4. Calculation of coordination and coordination variability

For each instant (i) during the normalized stance phase, the coupling angle (γi) was calculated based on the consecutive proximal segmental angles (θP(i), θP(i+1)) and consecutive distal segmental angles (θD, θD(i+1)) according to Eqs. (1) and (2):

\[ γ_i = \text{Atan} \left( \frac{θ_D(i+1) - θ_D(i)}{θ_P(i+1) - θ_P(i)} \right) \]

\[ γ_i = \frac{180}{π} \left( θ_P(i+1) - θ_P(i) \right) \]

\[ γ_i = \frac{180}{π} \left( θ_P(i) - θ_P(i+1) \right) \]

The following conditions (3) were applied:

\[ \text{if } i_p = 90 \text{ and } i_D = 0 \text{ then } γ_i = 0 \]

\[ \text{if } i_p = -90 \text{ and } i_D = 0 \text{ then } γ_i = 180 \]

\[ \text{if } i_p = 0 \text{ and } i_D = -90 \text{ then } γ_i = -180 \]

\[ \text{if } i_p = 0 \text{ and } i_D = 90 \text{ then } γ_i = 0 \]

Coupling angle (γi) was corrected to present a value between 0° and 360° according to (4) (Sparrow, Donovan, van Emmerik, & Barry, 1987).
Due to directional nature of coupling angle, the average coupling angle ($\gamma_i$) were calculated based on the average horizontal ($x_i$) and vertical ($y_i$) components at each instant using circular statistics (Hamill, Haddad, & McDermott, 2000).

$$x_i = \frac{1}{n} \sum_{i=1}^{n} \cos \gamma_i$$  \hspace{1cm} (5) \\
$$y_i = \frac{1}{n} \sum_{i=1}^{n} \sin \gamma_i$$  \hspace{1cm} (6)

The following (7) were applied to correct the average coupling angle ($\overline{\gamma}$) to present a value between 0° and 360°.

$$\overline{\gamma} = \begin{cases} 
\text{Atan} \left( \frac{y}{x} \right) \frac{180}{\pi} & x_i > 0, y_i > 0 \\
\text{Atan} \left( \frac{y}{x} \right) \frac{180}{\pi} + 180 & x_i < 0, y_i > 0 \\
\text{Atan} \left( \frac{y}{x} \right) \frac{180}{\pi} + 360 & x_i > 0, y_i < 0 \\
90 & x_i = 0, y_i > 0 \\
-90 & x_i = 0, y_i < 0 \\
\text{indefined} & x_i = 0, y_i = 0 
\end{cases}$$  \hspace{1cm} (7)

The length of average coupling angle $\tau$ was calculated according to (8)

$$\tau = \sqrt{x_i^2 + y_i^2}$$  \hspace{1cm} (8)

Coupling angle variability (CAV) was calculated according to (9)

$$CAV = \sqrt{2(1 - \overline{\gamma})} \frac{180}{\pi}$$  \hspace{1cm} (9)

Figs. 2 and 3 illustrate what a coupling angle indicates when it falls onto each of the quadrants, which is necessary for interpreting the results of Tables 1 and 3, respectively.

2.5. Protocol

All subjects wore the same neutral shoe model at both baseline and post-test measurements (New Balance 759, USA). Custom-made medial arch support FOs (Ethylene vinyl acetate and microcellular rubber were used to fabricate the FOs) were applied to produce the negative impression of the foot held in the subtalar joint neutral position in EG (Fig. 4A). FOs peak longitudinal height of the mid-foot arch was 25 mm in the EG while 2-mm-thick insoles (flat) were used for the CG (made of polyester resin as a sham condition) (Fig. 4B).

In the baseline assessment, subjects were asked to walk with standard shoes and without FOs. After the pre-tests, each subject was asked to wear FOs (EG) and insoles (CG) during daily activities for 4 months. During the intervention period, subjects were instructed to progressively increase the using time of their FOs (EG) and insoles (CG). On the first day, subjects were asked to confine the using time to one hour in order to become familiarized. On every following day, the using time was increased by one hour until subjects eventually wore the FOs (EG)/insoles (CG) for the full day (Moisan & Cantin, 2016). Subjects were instructed to complete a log in which the using time of FOs/insoles was quantified on every day. Post-tests were conducted at the same time of day and in the similar test sequence as the baseline tests.

2.6. Statistical analysis

Data are presented as group mean values and standard deviations. A Watson-Williams F-test was used to determine significant differences in between-group baseline values (pre-test values). Also, within-group comparisons (comparison between pre and post-test values of each group) were done by using a paired Watson-Williams test for circular variables. These analyses were performed using Oriana software version 3.21(Wales, UK). Coordination variability between and within groups was also compared using the independent t-test and paired t-test, respectively. Following Nakagawa (Nakagawa, 2004), we did not perform Bonferroni corrections for multiple comparisons because the correction would substantially reduce statistical power. We calculated the effect size for each significant result and quantified it using Cohen’s $d$ (Cohen Jacob, 1988). Accordingly, $d < 0.50$ indicate small effects, $0.50 \leq d < 0.80$ indicate medium effects, and $d \geq 0.80$ indicate large effects. The analysis was performed using SPSS (version 22). The significance level was set at $p < .05$. 

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Fig. 2. Relationship between phase angle in each quadrant and distal-proximal joints' relative motion. A coupling angle of 0° or 180° indicates distal joint motion without proximal joint motion. A coupling angle of 90° or 270° indicates proximal joint motion without distal joint motion. A vector angle of 45°, 135°, 225°, and 315° indicates equal relative motion between the proximal and distal joints.

Fig. 3. Relationship between phase angle in each quadrant and relative motion of ankle inversion/eversion and knee internal/external rotation. A vector angle of 0° or 180° indicates ankle motion without knee motion. A vector angle of 90° or 270° indicates knee motion without ankle motion. A vector angle of 45°, 135°, 225°, and 315° indicates equal relative motion between the knee and ankle.
Table 1
Experimental and control groups’ coupling angle (mean ± standard deviation) in each sub-phase of the stance. The sub-phases are categorized into the following gait events: loading response (LR; 0–15% of gait cycle), mid stance (MS; 15–45% of gait cycle), and push-off (PO; 45–60% of gait cycle). The coordination pattern classification is divided into the following categories: proximal phase (PP), distal phase (DP), in-phase (IP), anti-phase (AP). Unit for the values is degrees.

| Joints Coordination | Sub-phase | EG (n = 15) | p-value (Between Pre & post- Test of EG) | CG (n = 15) | p-value (Between Pre & post- Test of CG) | p-value (Between the groups Pre-Test) | p-value (Between the groups Post-Test) |
|---------------------|-----------|-------------|------------------------------------------|-------------|----------------------------------------|-------------------------------------|--------------------------------------|
|                     |           | Pre-Test    | Post-Test                                | Pre-test    | Post-test                              | Pre-test                           | Post-test                           |
| Ankle-Knee          | Sagittal  | LR          | 69.6 ± 20.6 (PP)                         | 82.5 ± 14.7 (PP) | 0.084                                  | 69.7 ± 20.3 (PP) 71.8 ± 11.2 (PP) | 0.429 0.521 0.482                   |
|                     |           | MS          | 337.8 ± 9.1 (DP)                         | 337.2 ± 4.8 (AP) | 0.519                                  | 341.3 ± 5.5 (DP) 338.5 ± 7.5 (DP) | 0.319 0.624 0.393                   |
|                     |           | PO          | 111.6 ± 3.1 (PP)                         | 110.3 ± 2.8 (PP) | 0.781                                  | 111.7 ± 3.1 (PP) 113.1 ± 2.7 (AP) | 0.351 0.818 0.212                   |
| Ankle-Knee          | Sagittal  | LR          | 103.2 ± 13.6 (PP)                        | 108.7 ± 16.4 (PP) | 0.941                                  | 100.9 ± 14.8 (PP) 103.6 ± 10.7 (PP) | 0.902 0.912 0.490                   |
|                     |           | MS          | 271.1 ± 29.3 (PP)                        | 284.5 ± 37.5 (PP) | 0.518                                  | 269.3 ± 20.6 (PP) 271.1 ± 22.9 (PP) | 0.427 0.319 0.202                   |
|                     |           | PO          | 101.5 ± 18.4 (PP)                        | 105.6 ± 26.9 (PP) | 0.905                                  | 103.4 ± 13.5 (PP) 102.4 ± 16.1 (PP) | 0.953 0.992 0.956                   |
| Ankle-Knee          | Transverse| LR          | 140.8 ± 7.1 (AP)                         | 135.6 ± 5.3 (AP) | 0.028                                  | 134.8 ± 2.4 (AP) 136.2 ± 2.3 (AP) | 0.745 0.861 0.425                   |
|                     |           | MS          | 17.4 ± 7.2 (DP)                          | 12.6 ± 5.1 (AP) | 0.047                                  | 16.1 ± 5.4 (AP) 13.5 ± 4.1 (DP) | 0.285 0.096 0.073                   |
| Ankle-Hip           | Sagittal  | LR          | 272.2 ± 20.6 (PP)                        | 254.1 ± 13.3 (PP) | 0.008                                  | 269.8 ± 24.1 (PP) 263.7 ± 11.8 (PP) | 0.278 0.163 0.182                   |
|                     |           | MS          | 299.7 ± 4.6 (AP)                         | 299.3 ± 3.6 (AP) | 0.711                                  | 301.5 ± 7.4 (AP) 301.1 ± 6.6 (AP) | 0.815 0.824 0.928                   |
|                     |           | PO          | 136.6 ± 7.5 (AP)                         | 139.4 ± 9.6 (PP) | 0.592                                  | 136.1 ± 8.9 (AP) 138.3 ± 7.9 (AP) | 0.449 0.638 0.510                   |
| Ankle-Hip           | Sagittal  | LR          | 106.9 ± 14.4 (PP)                        | 146.8 ± 39.7 (AP) | 0.002                                  | 104.5 ± 13.8 (PP) 103.5 ± 12.5 (PP) | 0.817 0.933 0.923                   |
|                     |           | MS          | 221.1 ± 101.4 (IP)                       | 156.9 ± 130.4 (IP) | 0.010                                  | 216.9 ± 108.6 (IP) 235.6 ± 95.8 (IP) | 0.194 0.085 0.007                   |
|                     |           | PO          | 262.8 ± 12.2 (IP)                        | 262.7 ± 17.8 (PP) | 0.989                                  | 257.2 ± 11.4 (PP) 260.4 ± 12.1 (PP) | 0.587 0.451 0.299                   |
| Ankle-Hip           | Transverse| LR          | 219.8 ± 4.3 (IP)                         | 212.1 ± 5.2 (IP) | 0.598                                  | 221.4 ± 4.5 (IP) 218.8 ± 3.7 (IP) | 0.456 0.635 0.273                   |
|                     |           | MS          | 35.2 ± 89.9 (IP)                         | 348.1 ± 4.3 (IP) | 0.068                                  | 36.8 ± 89.6 (IP) 11.7 ± 7.3 (DP) | 0.717 0.389 0.356                   |
| Knee-Hip            | Sagittal  | LR          | 317.3 ± 9.4 (AP)                         | 320.0 ± 8.6 (AP) | 0.951                                  | 317.7 ± 10.1 (AP) 316.4 ± 5.9 (AP) | 0.501 0.476 0.196                   |
|                     |           | MS          | 265.3 ± 4.8 (PP)                         | 263.9 ± 3.1 (PP) | 0.710                                  | 266.2 ± 4.5 (PP) 265.1 ± 4.0 (AP) | 0.795 0.866 0.555                   |
|                     |           | PO          | 21.8 ± 4.1 (IP)                          | 18.0 ± 2.9 (DP) | 0.006                                  | 21.4 ± 3.9 (IP) 21.2 ± 3.8 (DP) | 0.360 0.492 0.291                   |
| Knee-Hip            | Sagittal  | LR          | 35.5 ± 8.9 (IP)                          | 116.0 ± 149.7 (AP) | 0.010                                  | 36.7 ± 10.1 (IP) 35.3 ± 8.2 (IP) | 0.081 0.093 0.004                   |
|                     |           | MS          | 192.4 ± 16.8 (DP)                        | 174.3 ± 19.8 (DP) | 0.012                                  | 190.0 ± 15.4 (DP) 191.6 ± 14.6 (IP) | 0.927 0.984 0.906                   |
|                     |           | PO          | 303.9 ± 7.6 (AP)                         | 298.2 ± 7.4 (AP) | 0.928                                  | 304.0 ± 5.2 (AP) 301.8 ± 6.4 (AP) | 0.981 0.956 0.933                   |
| Knee-Hip            | Sagittal  | LR          | 314.1 ± 14.7 (AP)                        | 328.8 ± 14.1 (AP) | 0.871                                  | 322.6 ± 5.7 (AP) 324.3 ± 6.1 (AP) | 0.739 0.829 0.600                   |
|                     |           | MS          | 60.9 ± 80.3 (IP)                         | 281.5 ± 140.9 (IP) | 0.159                                  | 68.1 ± 79.7 (IP) 64.3 ± 78.6 (IP) | 0.268 0.127 0.071                   |
|                     |           | PO          | 173.6 ± 7.6 (DP)                         | 172.2 ± 1.1 (DP) | 0.928                                  | 166.8 ± 19.3 (DP) 169.9 ± 12.2 (DP) | 0.930 0.978 0.917                   |

⁎ Significant difference between the results of pre and post-test within the experimental group (bolded values).

⁎⁎ Significant difference between the results of experimental and control groups during post-test assessment (bolded values).
3. Results

3.1. Joint coupling analysis

There were no statistically significant differences in baseline (pre-test) values between the experimental and control groups ($p > .05$). Table 1 compares the mean coupling angles within each group as well as between the groups during each sub-phase of the stance. Paired Watson-Williams test showed no statistically significant difference between the results obtained from the subjects in CG before and after four months ($p > .05$) (see Table 3). However, foot orthotic therapy changed the relative motion of joints, as follows:

3.1.1. Ankle-knee coordination

Significant differences for the ankle-knee coordination occurred only in the transverse plane. During loading response, the mean coupling angle of EG in pre-test was $140.8 \pm 7.1^\circ$, and it was $135.6 \pm 5.3^\circ$ in post-test ($p = .028; d = 0.84$). This indicates that after four months, the subjects in EG showed an absolutely anti-phase pattern for ankle/knee coordination.

During mid-stance, the EG’s coupling angle in the post-test was $17.4 \pm 7.2^\circ$, and it was $12.6 \pm 5.1^\circ$ in the pre-test ($p = .047; d = 0.78$). This indicates that even though the subjects in EG moved both ankle and knee into internal rotation, they showed more ankle internal rotation during post-test compared to pre-test.

During push-off, the EG’s coupling angle in the post-test ($240.8 \pm 7.6^\circ$) was significantly different from the EG’s coupling angle in the pre-test ($224.5 \pm 15.9^\circ$) ($p < 0.001; d = 1.38$), as well as form the post-test of CG ($222.7 \pm 14.8^\circ$) ($p < 0.001; d = 1.61$). This indicates that after four months, the subjects in EG indicated more knee motion relative to ankle motion in the post-test assessment.

3.1.2. Ankle-hip coordination

In the sagittal plane during loading response, the mean coupling angle of the EG was $272.2 \pm 20.6^\circ$ in the pre-test which was close to $270^\circ$ indicating a hip-dominant coupling strategy, while it was $254.1 \pm 13.3^\circ$ during post-test demonstrating that the subjects moved the ankle into plantar flexion while moving the hip into extension ($p = .008; d = 1.06$).

In the frontal plane during loading response, the mean coupling angle of the EG in the post-test was $146.8 \pm 39.7^\circ$ and it was $106.9 \pm 14.4^\circ$ in the pre-test ($p = .002; d = 1.47$). This indicates that although the subjects moved the ankle into eversion while moving the hip into adduction, they had more ankle motion relative to hip motion during post-test. Furthermore, at mid-stance, although the subjects of the EG moved the ankle into eversion while moving the hip into abduction during pre-test ($221.1 \pm 101.4^\circ$), they experienced ankle eversion and hip adduction during post-test ($156.9 \pm 130.4^\circ$) ($p = .01; d = 0.55$). Similarly, such a significant difference was also seen between EG ($156.9 \pm 130.4^\circ$) and CG ($235.6 \pm 95.8^\circ$) during post-test assessment ($p = .007; d = 0.69$).

In the transverse plane during push-off, although the participants of EG experienced ankle external rotation and hip internal rotation in both pre-test ($168.3 \pm 8.5^\circ$) and post-test ($161.2 \pm 8.7^\circ$), they showed a more anti-phase pattern during post-test ($p = .034; d = 0.82$).

3.1.3. Knee-hip coordination

In the sagittal plane during push-off, the mean coupling angle of the EG in the post-test ($18 \pm 2.9^\circ$) was significantly different from that during the pre-test ($21.8 \pm 4.1^\circ$) ($p = .006; d = 1.08$). This indicates that although the subjects moved both knee and hip

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Fig. 4. (A) illustrates an arch support foot orthoses that was applied in the experimental group. (B) shows a flat 2-mm-thick insole used as sham condition in the control group.
joints into flexion, they had more knee motion relative to hip motion during post-test.

In the frontal plane during loading response, the subjects of the EG moved the both knee and hip into adduction (35.5 ± 8.9°) in the pre-test assessment; however, they showed an anti-phase pattern indicating knee abduction and hip adduction in the post-test. Similarly, such a significant difference was also observed between EG (116.0 ± 149.7°) and CG (35.3 ± 8.2°) during post-test assessment (p = .004; d = 1.02).

In the frontal plane during mid-stance, the coupling angle differed significantly from 192.4 ± 16.8° in the pre-test to 174.3 ± 19.8° in the post-test (p = .012; d = 0.98). This indicated that although the subjects moved the both hip and knee into abduction in pre-test, they showed a trend towards hip adduction during post-test assessment.

### 3.2. Analysis of joint coordination variability

There were no statistically significant differences in baseline (pre-test) CAV values between the experimental and control groups (p > .05). Table 2 compares CAV values within each group as well as between the groups during each sub-phase of the stance. No significant difference was observed in the CG in the post-test relative to pre-test. Compared to the pre-test, the mean CAV of EG in the post-test decreased for the ankle-knee in the sagittal plane during loading response (p = .016; d = 2.86), for the ankle-hip in the sagittal plane during mid-stance (p = .046; d = 0.8), and for the ankle-hip in the frontal plane during push-off (p = .003; d = 1.16).

In contrast, CAV were greater in the EG for the ankle-hip in sagittal plane during push-off (p = .005; d = 1.12), ankle-hip coordination in transverse plane during loading response (p = .003; d = 1.32) and mid-stance (p = .017; d = 0.93), as well as knee-hip in transverse plane during loading response (p = .014; d = 1.14) and mid-stance (p = .001; d = 1.48) in the post-test compared to the pre-test.

In terms of between-groups comparisons during post-test assessment, CAV were greater in the EG for the ankle-hip in sagittal plane during push-off (p = .006; d = 1.25), ankle-hip in transverse plane during mid-stance (p = .011; d = 1.23), knee-hip in transverse plane during loading response (p = .013; d = 1.81) and mid-stance (p = .001; d = 1.84) compared to the CG. However, the
Table 3
Mean ankle inversion/eversion and knee internal/external rotation joint coupling angle and its variability for the experimental and control groups during each sub-phase of the stance. The coordination pattern classification is divided into the following categories: proximal phase (PP), distal phase (DP), in-phase (IP), anti-phase (AP).

| Sub-phase | Ankle inversion/eversion & knee internal/external rotation | EG (n = 15) | Pre-Test | Post-Test | p-value (Between Pre & Post-Test of EG) | CG (n = 15) | Pre-Test | Post-Test | p-value (Between Pre & Post-Test of CG) | p-value (Between the groups Pre-Test) | p-value (Between the groups Post-Test) |
|-----------|----------------------------------------------------------|-------------|----------|----------|----------------------------------------|-------------|----------|----------|----------------------------------------|--------------------------------------|--------------------------------------|
| Joint Coordination | LR | 106.3 ± 15.8 (PP) | 109.6 ± 11.1 (PP) | 0.741 | 110.7 ± 15.6 (PP) | 105.3 ± 19.4 (PP) | 0.645 | 0.712 | 0.286 |
| | MS | 88.7 ± 49.2 (PP) | 58.9 ± 46.8 (IP) | 0.038 | 83.8 ± 24.5 (PP) | 76.1 ± 33.4 (PP) | 0.312 | 0.297 | 0.191 |
| | PO | 267.3 ± 26.5 (PP) | 260.6 ± 14.9 (PP) | 0.985 | 256.2 ± 34.1 (PP) | 268.9 ± 18.3 (PP) | 0.967 | 0.835 | 0.697 |
| Coordination Variability | LR | 41.3 ± 12.2 | 27.5 ± 7.9 | 0.082 | 38.2 ± 17.2 | 32.7 ± 19.4 | 0.113 | 0.216 | 0.107 |
| | MS | 65.9 ± 21.1 | 65.1 ± 6.2 | 0.996 | 68.1 ± 15.6 | 61.3 ± 18.5 | 0.943 | 0.562 | 0.210 |
| | PO | 29.8 ± 10.1* | 15.9 ± 9.3* | 0.028 | 23.9 ± 17.3 | 27.1 ± 15.2 | 0.459 | 0.352 | 0.096 |

* Significant difference between the results of pre- and post-test within the experimental group (bolded values).
mean CAV of EG decreased for the ankle-knee in the sagittal plane during loading response \( (p = .014; d = 0.87) \).

Table 3 describes pre- and post-intervention results for the ankle inversion/eversion and knee internal/external rotation joint coupling angle and its coordination variability for the experimental and control groups during each sub-phase of the stance. For the EG during mid-stance, results showed a pattern indicating more ankle inversion at the post-test relative to the pre-test \( (p = .038) \). The mean CAV of EG in post-test assessment compared to pre-test decreased for the ankle inversion/eversion and knee internal/external rotation during push-off \( (p = .028) \). However, there were no significant differences between the EG and CG for ankle inversion/ eversion and knee internal/external rotation angle and its variability during pre- and post-test assessments \( (p > 0.05) \).

4. Discussion

Understanding the inter-joint coordination and coordination variability is important to clarify the mechanism of abnormal gait and occurrence of injury. In this study, we quantified differences in lower inter-joint coordination and its variability before and after four months wearing arch support FOs in children with flexible flat feet.

4.1. Analysis of inter-joint coordination

4.1.1. Ankle-knee coordination

Our results for ankle in/eversion-knee in/external rotation joint coordination angle of EG demonstrated that during mid-stance subjects showed a pattern indicating more ankle inversion during post-test relative to pre-test. A previous study reported that lower rearfoot in/eversion-tibia in/external rotation coordination in individuals with pronated foot is mainly due to having greater tibia rotation relative to healthy subjects (McClay & Manal, 1997). FOs are typically made to control rearfoot eversion; hence, they will probably increase the proportion of inversion to knee internal rotation and therefore change their relative coordination. Our results confirmed this issue and showed that ankle in/eversion-knee in/external rotation changed to an in-phase pattern (59°) in the post-test compared to a clear knee internal rotation pattern (89°) in the pre-test. It is reported that individuals with flat feet who have a greater ankle in/eversion-knee in/external ratio, with a rearfoot eversion dominancy, show a higher risk of knee injuries (D. S. Williams, McClay, & Hamill, 2001). A study evaluating the immediate effect of standard orthoses on the rearfoot eversion/tibia internal rotation coordination of healthy runners found increased rearfoot eversion/tibia internal rotation excursion coordination mainly due to reduced tibia internal rotation (Nawoczenski et al., 1995). However, they did not determine the effects of long-term wearing of FOs, so it is difficult to compare their results with those of the present study. Also, another study did not report any significant differences in the joint coupling angle or variability between rearfoot and knee when subjects ran in the FOs condition compared to the no device condition (Ferber, Davis, & Williams, 2005).

In the EG, the values of all 3 sub phases for transverse plane ankle-knee coordination differed significantly in the post-test relative to pre-test. It is known that the normal function for transverse plane ankle-knee coordination during landing response occurs when ankle and knee joints cooperate in the anti-phase pattern (Ferber et al., 2005). This is supported by the result of the current study in which an absolutely anti-phase pattern was shown during landing response in the post-test. Transverse plane ankle-knee coordination in the EG during push-off increased in the post-test compared to the pre-test as well as compared to the post-test of CG. This expresses although this motion coordination is still in the in-phase pattern, it moves with the dominance of knee rotation.

During mid-stance, the subjects moved both ankle and knee into internal rotation, they showed more ankle motion relative to knee motion during post-test assessment. This leads to improved shock absorption and reducing forces accumulated on the foot (Perry & Lafontaine, 1995; Pratt, 1989). Previous studies reported that FOs devices may be very effective in reducing knee pain (Janice J Eng & Pierrynowski, 1993). This reduction in the knee pain after orthotic therapy may be in some part due to changes in ankle-knee coupling angles observed in the present study. However, further study is needed to confirm this issue.

4.1.2. Ankle-hip coordination

In the frontal plane during landing response, the EG's coupling angle in the post-test was significantly different from that during the pre-test. The results indicated that although the subjects of EG moved the ankle into eversion while moving the hip into adduction, they had more ankle motion relative to hip motion during the post-test. Moreover, during mid-stance, although the subjects moved the ankle into eversion while moving the hip into abduction during pre-test, they showed ankle eversion and hip adduction during post-test (an anti-phase pattern). An increase in closed-chain hip adduction (lateral shift of the pelvis) could decrease the risk of eversion-related injuries because this could move the center of mass (COM) further towards the lateral border of the foot to push the ankle into inversion (Yen, Chui, Corkery, Allen, & Cloonan, 2017). This is especially the case when the ankle eversion is relatively high to bring the joint into a more everted position. This suggests that enhancing the quality of arch support FOs to control excessive subtalar eversion may be important in the gait rehabilitation of individuals with flat feet.

Also, in the transverse plane during push-off, although the participants experienced ankle external rotation and hip external rotation in both pre-test and post-test, they showed a more in-phase pattern during post-test. It has been suggested that the hip and the subtalar joint work synergistically to control foot placement and COM during walking (MacKinnon & Winter, 1993), and proper control of the relative position between the COM and the foot plays an important role in ankle injuries prevention.

4.1.3. Knee-hip coordination

Our results showed an anti-phase pattern for frontal plane knee-hip coordination in the post-test of EG during landing response indicating knee abduction and hip adduction while CG showed an in-phase pattern in both pre-test and post-test. This is in line with
previous studies showing knee abduction and hip adduction during the loading response of walking (Koshino et al., 2017). In the sagittal plane during push-off, the mean coupling angle of the EG showed more knee motion relative to hip motion in the post-test than that in the pre-test. This indeed indicates the dominancy of knee flexion relative to hip flexion during push-off in the post-test.

4.2. Analysis of inter-joint coordination variability

4.2.1. Ankle-knee coordination variability

Decreased ankle in/eversion-knee in/external rotation CAV during push-off is shown for EG in the post-test than pre-test. Ankle in/eversion-knee in/external rotation CAV is important in impact force absorption (Floría, Sánchez-Sixto, Ferber, & Harrison, 2018). A study reported that custom FOs may have a role in the maintenance of coordination variability of the ankle in/eversion-knee in/external rotation (MacLean et al., 2010). Timing interruption (i.e. coupling angle variability) in the ankle in/eversion-knee in/external rotation can link the ankle eversion to the potential knee injury risk (DeLeo, Diers, Ferber, & Davis, 2004). Since shock absorption occurs in the first stance phase, decreased CAV at loading response in the post-test, despite that it is not significant, may increase stress on the knee when using FO. In the current study, although decreased ankle in/eversion-knee in/external rotation CAV is shown at loading response and push-off, the same CAV is shown between post-test and pre-test at midstance. This implies that using FOs affects motor flexibility and leads to decrease in the flexibility of the ankle in/eversion in the first and late stance phase.

Decreased sagittal plane ankle-knee CAV during loading response is shown for the EG in the post-test compared to the pre-test. No significant difference was found for ankle-knee CAV in the other planes between post-test and pre-test. Using orthotics may restrict dorsiflexion motions in the affected foot, as previous studies reported reduced peak dorsiflexion and range of motion by using orthotics (Donoghue, Harrison, Laxton, & Jones, 2008). This restriction causes the knee to go into hyperextension and potential compensatory motions in the trunk (L. Perry & Burnfield, 2010). These changes during gait may be the reason for reducing ankle-knee coordination variability in the sagittal plane for the EG.

4.2.2. Ankle-hip coordination variability

Reduced frontal plane ankle-hip CAV during push-off is shown for the EG in the post-test than pre-test. Ankle and hip joints should work collaboratively to control foot placement and center of gravity during gait (DA A. Winter, 1995). Two factors may affect joint coordination variability: 1, relative motion and 2, range of motion (DeLeo et al., 2004). Therefore, reduced CAV might be a result of using FOs in which either relative motion or range of motion of ankle is reduced in the frontal and sagittal planes.

The results show increased transverse plane ankle-hip CAV at both loading response and mid-stance for EG in the post-test than pre-test. Also, sagittal plane ankle-hip CAV increased during push-off in the post-test for EG relative to pre-test of EG and post-test of CG. It is suggested that greater CAV is associated with greater flexibility of motor control to adapt itself with perturbations and sudden changes occurring during locomotion (Hafer, Brown, & Boyer, 2017; Silvernail, Boyer, Rohr, Brüggemann, & Hamill, 2015). Therefore, greater CAV may reduce repetitive stresses on lower limb joints and tissues. Previous studies reported that decreased ankle-hip CAV is associated with lower limb injuries such as ankle sprain (Yen et al., 2017), patellofemoral pain syndrome (Hamill et al., 1999), and iliotibial band syndrome (Miller et al., 2008). However, it is difficult to compare the results of these studies with those of the current study due to the different populations and different methods for calculating CAV (Continuous relative phase versus vector coding).

4.2.3. Knee-hip coordination variability

The results show increased transverse plane knee-hip CAV at both loading response and mid-stance for EG in the post-test relative to pre-test of EG and post-test of CG. However, no significant differences are shown for the sagittal and frontal plane knee–hip CAV between pre-test and post-test of the EG. Previous studies highlighted knee-hip coordination and its variability as an important factor for lower limb function (Powers, 2010; Willson & Davis, 2008). The current results stress that a four-month wearing FOs causes greater flexibility in knee-hip coordination in the transverse plane resulting in better functioning of lower extremity motions, impact force attenuation and consequently injury prevention (Powers, 2010). Although there is currently no research to compare the results of this study, this paper represents information regarding three-dimensional lower limb inter-joint coordination and its variability after long term use of FOs in male children with flexible flat feet during gait. These novel data may establish the baseline information for future studies in this area and can contribute to research design and sample size calculations for larger clinical trials involving children with flexible flat feet pathologies and other related clinical conditions.

Several researchers indicated that inter-joint coordination and its variability play a key role in maintaining dynamic balance as well as the adaptability required to adjust movements during gait (Chiu & Chou, 2013; van Emmerik, Ducharme, Amado, & Hamill, 2016). Therefore, our results may have implications to using FOs complementary to training programs to improve inter-joint coordination and/or its variability in children with flexible flat feet.

4.3. Limitations and recommendations for future studies

This study exhibits some methodological limitations that warrant discussion. Only boys aged 8–12 years were enrolled in this study. Therefore, our findings cannot be generalized to younger and older boys due to growth and maturation and to girls due to differences in biomechanical gait characteristics. Another potential limitation of this study refers to the use of the Plug-in Gait model. It has previously been speculated that this model might not be precise and accurate enough to quantify ankle joint kinematics. Therefore, it is recommended that future studies apply 6 degrees of freedom marker set models instead of simple Plug-in-Gait marker.
sets.

5. Conclusion

The findings of this study showed that four months wearing arch support FOs can alter inter-joint coordination and its variability in children with flexible flat feet. Our findings suggest that long-term use of arch support FOs in children with flat feet is associated with an in-phase pattern in the ankle in/eversion-knee in/external rotation coupling. Moreover, results show increased CAV for most of the parameters after 4-month wearing FOs. These findings provided significant implications for the understanding of how the long-term use of arch support FOs might be effective to alter lower limb inter-joint coordination and coordination variability in children with flexible flat feet during walking. Since there is no data about children showing a standard coordination and coordination variability magnitude for the most parameters evaluated in this study, we cannot reach a concrete conclusion for our results. It is, therefore, an opportunity for future studies to investigate lower limb joint coordination and its variability between children with flat feet and normal foot. Taken together, the insights gained from this study may be of assistance in applying FOs for children with flexible flat feet and improving correction exercises strategies.

Declaration of Competing Interest

The authors declare that they have no conflict of interest relating to the material presented in this article.

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