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Muscle fatigue during a short walking exercise in children with cerebral palsy who walk in a crouch gait

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ABSTRACT

Background: A deterioration of crouch gait was found in a group of children with cerebral palsy (CP) after a short walking exercise. The increased knee flexion reported after a continuous walk could be related with muscle fatigue and muscle strength.
Aim: Does muscle fatigue appears at the end of a walking exercise in children with CP who walk in a crouch gait?
Methods: Eleven children with cerebral palsy (GMFCS I to III) who walk in a crouch gait were included. Isometric muscle strength was assessed using a handheld dynamometer. Children were asked to walk for 6 min at comfortable speed. Spatio-temporal, kinematic and electromyographic (EMG) measurements were recorded at the first and the last minute of the 6-minute walking exercise. Muscle fatigue was evaluated using the shift of EMG signals median frequency.
Results: There was no significant difference in walking speed, cadence, and step length at the end of the 6mwe. Maximal and mean anterior pelvic tilt decreased and knee flexion increased ($p < 0.05$). Rectus femoris EMG median frequency decreased ($p < 0.05$). The median frequency in other muscles did not decrease significantly. Greater hip extensor strength was associated with lesser knee flexion at the end of the 6-minute walking exercise ($p < 0.05$).
Significance: The increase in knee flexion at the end of the 6-minute walking exercise can be explained by muscle fatigue found in rectus femoris. Hip extensor strength can limit the deterioration of crouch gait after a 6-minute walking exercise representative of daily activities.

1. Introduction

Cerebral palsy (CP) represents a group of posture and movement disorders that results from non-progressive lesions on a developing brain [1]. Children with CP have impaired motor control, abnormal muscle tone, and muscle weakness [2], which lead to functional disabilities, including impaired walking. Crouch gait, characterized by excessive hip and knee flexion during the stance phase of gait, is one of the most common gait pattern in spastic bilateral CP [3]. Such a walking pattern leads to excessive mechanical stress at hip and knee joints [4]. It also requires greater muscle work to walk, which increases with crouch severity [5] and may expose to early fatigue during short walking exercise.

To date, very few studies have investigated gait adaptations during a continuous walking exercise representative of daily life in children with CP [6,7]. In children who walk in a crouch gait, a kinematic study showed that a short walking exercise exacerbates crouch gait pattern [6]. Indeed, children had a greater knee and ankle flexion at the end of a 6-minute walking exercise performed at comfortable speed [6]. Thereby, this study suggested that, within short walking distance,

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children who walk in a crouch gait have a deterioration of their gait pattern due to muscle fatigue. However, no fatigue indicator was reported to support this hypothesis.

Recently, Eken et al. [7] reported muscle fatigue in lower leg muscles in children with CP at the end of a 5-min walking exercise [7]. The authors used the median frequency (MDF) of electromyography (EMG) signals as muscle fatigue indicator, which is expected to shift toward lower frequencies in presence of muscle fatigue [8,9]. However, the muscle fatigue found during gait by Eken et al. [7] was not reported along with kinematic measurements [7] that would have help to interpret the impact of muscle fatigue on gait. Moreover, this study highlighted muscle fatigue at lower leg muscles in a group of children with moderate functional abilities, i.e. mostly Gross Motor Function Classification System (GMFCS) level I, who walked with various gait patterns. Considering that fatigue is particularly reported by children with CP with poor locomotion capacity, namely children with GMFCS level II and III [10], it would be relevant to evaluate the impact of a walking exercise on muscle fatigue in this group of children. Finally, in regards to the large variability of gait patterns in children with CP [11], and considering that muscle work can greatly differ between gait patterns, it is essential to address this question for a specific gait.

Children with CP who walk in a crouch gait represent a homogeneous group of children with important muscle weakness [5] that affects walking capacities. Evaluating the presence of muscle fatigue during a walking exercise corresponding to daily life activities in these children may help to precise therapeutic intervention. The objective of the present study was to investigate kinematic changes and muscle fatigue in children who walk in a crouch gait at the end of a walking exercise, and to study the correlation between changes in crouch gait and muscle strength. It was hypothesized that increased knee flexion and muscle fatigue at the knee extensors would appear at the end of the exercise. It was also expected that children with lower hip and knee extensor strength would be more prone to walking pattern modifications.

2. Methods

2.1. Participants

The study included 11 children with bilateral spastic CP (GMFCS I: 2; GMFCS II: 5; GMFCS III: 4) who undergone an instrumented gait analysis at the *Gait lab Montreal (LAM-CRME)* of Sainte-Justine Hospital between 2013 and 2017. Inclusion criteria were a diagnosis of bilateral spastic CP, the ability to walk for 6 min with or without walking aids, and a knee flexion greater than 15° throughout the stance phase of gait (Supplementary Fig. 1). Children were excluded if they had a surgical intervention in the last 12 months, botulinum toxin injections in the last 6 months, an inability to walk without orthosis, and/or inadequate kinematic or EMG data from the walking exercise during the gait analysis. All children's characteristics are reported in Table 1. Consent and assent were obtained from all children's parents and children, respectively. This study was approved by the Research Ethics Board of Sainte-Justine Hospital.

2.2. Experimental protocol and data collection

2.2.1. Muscle strength

Before gait analysis, children performed hip and knee maximal isometric voluntary contractions to assess their muscle strength. Hip and knee flexor/extensor and hip abductor muscle force of either sides were measured 3 times by an experienced assessor using a handheld dynamometer (Lafayette Instruments, Lafayette, USA). According to Eek et al. [12], the knee flexor/extensor muscle force were evaluated in sitting position, the hip flexor and abductor muscle force in supine position and the hip extensor muscle force in prone position [12]. The assessor held the device on the distal portion of the femur or shank, while an assistant ensure the stability of the proximal portion of the segment. Participants were asked to push "as hard as possible" for 6 s. Standardized verbal encouragements were provided during the test.

2.2.2. Instrumented gait analysis

2.2.2.1. Kinematic measurements. A set of 35 reflective markers were positioned on anatomical landmark based on the full body Plug-in Gait kinematic model [13]. A 12-camera motion capture system (T40x cameras, Vicon, Oxford, UK) was used to measure the 3D markers displacement with a sampling frequency of 100 Hz.

2.2.2.2. EMG measurements. The EMG of the rectus femoris (RF), vastus lateralis (VL), tibialis anterior (TA), semitendinosus (ST), gastrocnemius lateralis (GAL), and gluteus medius (GM) was recorded using the wireless FreeEMG300 system (BTS Bioengineering, Milano, Italy). After standard skin preparation, bipolar self-adhesive surface Ag/AgCl electrodes (Covidien Kendall, Mansfield, MA, USA) were placed on the muscles belly with a 20 mm inter-electrodes distance following SENIAM recommendations [14]. Sampling frequency was set to 1000 Hz.

2.2.2.3. 6-minute walking exercise (6mwe). Children were asked to walk barefoot continuously for 6 min around a 25-meter path. Unlike the standard 6-minute walk test [15], children were asked to walk at their self-selected speed. This exercise was considered as representative of daily walking distances [16]. Standardized encouragements were provided. The kinematic and EMG signals were recorded at each minute while the participant walked along the straight part of the path. Only data from the first and last minutes were used in the subsequent analysis.

Table 1
Children's characteristics.

Participants	Gender	Age (years)	GMFCS	Assistive device	Mass (kg)	Height (cm)
1	M	10	II	None	30.4	133.0
2	M	16	III	Cane	31.5	163.0
3	M	14	II	None	43.0	165.0
4	M	14	II	None	47.0	165.0
5	F	11	III	Walker	35.0	137.0
6	M	16	I	None	54.3	167.5
7	F	12	I	None	45.2	162.0
8	M	14	II	None	31.7	137.8
9	M	12	III	None	44.4	173.0
10	M	11	III	Cane	27.2	131.1
11	M	9	II	None	26.8	126.8
Mean (std)	**2F – 11M**	**13 (2)**	–	–	**37.9 (9.2)**	**151.0 (1)**

Abbreviations: F – Female; M – Male; GMFCS – Gross Motor Function Classification System.

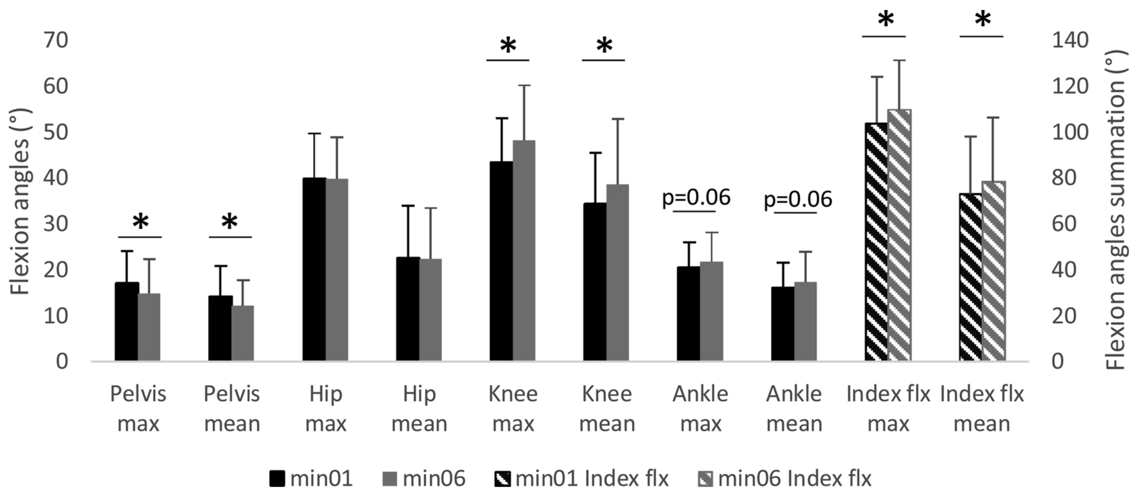

Fig. 1. Group average (+SD) maximal and mean flexion angles and flexion indexes at the first and sixth minute of the walking exercise. * Significant difference.

2.3. Data analysis

2.3.1. Muscle strength

The peak force reached during each maximal voluntary isometric contraction was recorded by the dynamometer and the two closest values were averaged. This force value was then multiplied by the lever arm length to obtain joint torque and normalized to body mass (Nm/kg).

2.3.2. Kinematics

Marker trajectories were analyzed using Nexus 1.8.5 (Vicon Motion Systems, Oxford, UK) and a custom-made Matlab (Mathworks, Natick, USA) script. The Vicon woltring routine was used to filter the 3-D marker trajectories [17]. The average of 2–5 gait cycles was used at each minute for analyses. The outcome measures were: (1) walking speed, step length, cadence; (2) mean and maximal pelvis, hip, knee and ankle joints angles in sagittal plane, (3) mean and maximal flexion indexes, corresponding to the summation of hip, knee and ankle mean and maximal flexion angles, respectively; and (4) mean and maximal center of mass (CoM) vertical trajectory normalized to body height. Only the kinematic data during the single limb stance phase were kept for analyses.

2.3.3. EMG median frequency

EMG data were analyzed using custom-made Matlab scripts [18]. EMG signals were 20–400 Hz bandpass filtered with a 4th order Butterworth filter. Thereafter, bandpass filtered EMG signals were rectified and 50 Hz low pass filtered to determine the EMG activation frames. The threshold was determined using EMG signals recorded during a relaxed state upright position. The 300 ms window with the smallest standard deviation (SD) was selected. Its mean added to 3SD was used to determine this activation threshold [19]. As EMG signals are non-stationary processes, wavelets analysis have been recommended to obtain the EMG MDF during EMG activation periods throughout the gait cycle [8,9]. A Morlet continuous wavelet transform was used (WavCrossSpec package [20], wave number: 7, frequency range: 1:400 Hz in 1 Hz steps) and the MDF was computed at each time-frame. Finally, EMG MDF was averaged during periods where EMG was above the activation threshold at the first and last minute of the 6mwe.

2.4. Statistical analysis

Both legs (22 limbs) were included in the statistical analysis. Normality of the distribution was evaluated using the skewness and kurtosis of the distribution. Depending on the normality of the distribution, a Student test or Wilcoxon signed rank test was used to compare kinematic and EMG MDF at the beginning and at the end of the walking exercise. Significance was set at $p \leq 0.05$, and corrected with false discovery rate (FDR) procedure for multiple comparisons ($q = 0.10$; FDR = 10%) [21]. The kinematic analysis included 11 children (22 limbs). Two children had no EMG data during the 6mwe. Therefore, the EMG analysis included 9 children (18 limbs). Moreover, due to EMG artifacts, the EMG analysis included various number of limbs depending on muscles, i.e. 14 limbs for the RF, TA and GAL; 10 limbs for the VL; 15 limbs for the GM; and 16 limbs for the ST.

Pearson's correlations was used to evaluate correlations between the significant kinematic changes, and initial extensor muscles strength. Kinematic changes corresponded to the difference between the sixth and first minute relative to the first minute.

3. Results

3.1. Kinematics

During the 6mwe, children walked a mean distance of 348 (± 116) m. Walking speed, cadence and step length did not change significantly between the first and the last minute of the walking exercise ($p > 0.05$, $q > 0.3$). Maximal and mean pelvic anterior tilt decreased ($p \leq 0.001$, $q = 0.01$) and knee maximal and mean flexion ($p < 0.01$, $q = 0.01$, and $q = 0.02$, respectively) increased significantly at the end of the 6mwe, as shown in Fig. 1. Maximal and mean ankle dorsiflexion increased, but did not reach significance ($p = 0.06$, $q = 0.16$). Maximal and mean flexion indexes increased significantly ($p < 0.05$, $q = 0.08$, and $q = 0.09$, respectively). Maximal angles values at the first minute and the variation of these angles is detailed in Supplementary Table 1.The mean normalized CoM vertical position decreased significantly at the end of the 6mwe (-0.36% of height, $p = 0.028$, $q = 0.096$).

3.2. Median frequency

The MDF of the RF decreased significantly between the first and sixth minute ($p = 0.002$, $q = 0.01$) (Fig. 2). Other muscles did not show significant modulation of MDF.

3.3. Correlations between kinematics and muscle strength

Hip extensor muscle strength was mildly correlated to changes in maximal knee flexion (Pearson's $r = -0.460$; Fig. 3) and changes in flexion index (Pearson's $r = -0.463$; Fig. 3). No significant correlation was found with knee extensor strength.

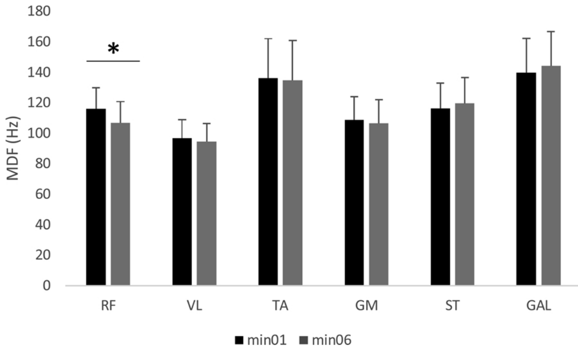

Fig. 2. Mean (+SD) median frequency at the first and sixth minute of the walking exercise. Abbreviations: RF – Rectus femoris; VL – Vastus lateralis; TA – Tibialis anterior ; GM – Gluteus medius; ST – Semitendinosus; GAL – Gastrocnemius lateralis. * Significant difference.

4. Discussion

The objectives of the present study were to evaluate crouch gait changes and muscle fatigue at the end of a continuous walking exercise in children with CP, and to report correlations between crouch gait changes and muscle strength. An increase in knee flexion, as well as a decrease in pelvic anterior tilt and in the CoM vertical position during walking were found. In addition, muscle fatigue at the RF was highlighted at the end of this exercise performed at self-selected walking speed. Finally, correlations were found between the increase in crouched posture and hip extensor strength, but not with knee extensor strength, as hypothesized.

During the last minute of the 6mwe, children had a greater knee flexion and ankle dorsiflexion ($p = 0.06$), as well as a decrease in CoM vertical position. These results are similar to those reported by Parent et al. [6] in a smaller group of children who walked with a crouch gait [6]. In the present study, hip flexion was not significantly increased. The reduction of pelvic anterior tilt, which decreases the hip flexion angle, explains this result. As this pelvic position has an impact on the inability to extend lower limbs [22], the decrease in pelvic anterior tilt may be interpreted as an adaptation to facilitate the extensor role of the gluteus maximus. In crouch gait, gluteus maximus plays a role to accelerate hip and knee towards extension [22]. Therefore, the decrease in pelvic anterior tilt can allow the gluteus maximus to compensate for fatigue at the knee extensor group. Another hypothesis for the posterior movement of the pelvis may be the alteration of the agonist-antagonist muscle balance due to fatigue of the RF. The RF has been highlighted as a major contributor of pelvis stabilization during hip flexion [23]. Therefore, fatigue at this muscle may have an impact on pelvic tilt.

Muscle fatigue in RF muscle, attested by a shift of the EMG MDF toward lower frequencies, was found along with the increase in crouched posture during walking at the end of the 6mwe. As walking in a crouch gait requires greater work from quadriceps muscles [5], muscle fatigue at the knee extensor muscles was anticipated at the end of such an exercise. Specifically, fatigue at VL muscle was more expected than fatigue at RF muscle, because it is the greater contributor to knee extension during single-limb stance in crouch gait [24]. However, muscle fatigue was only reported at the RF. It is interesting that muscle fatigue was only found in the biarticular muscle of quadriceps. Biarticular muscles are commonly used for controlling movement coordination [25]. The RF muscle fatigue in the present study may indicate a greater contribution of this biarticular muscle during continuous walking in crouch gait to control the forward acceleration and to help the VL to generate a vertical acceleration towards extension [26]. The difference in muscle fiber recruitment between VL and RF [27], suggesting a difference in muscle fiber type, may explained the early fatigue of the RF muscle. Indeed, the decrease in RF MDF only could be related to a lower prevalence of fatigue-resistant muscle fibers in this muscle compared to the VL [27]. Finally, one can argue that cross-talk between VL and RF muscles can occur, and would overestimate RF muscle activity with VL activity during gait [28,29]. However, in regards to RF and VL EMG patterns and due to the difference in RF and VL MDF variation, it is unlikely that cross-talk has influenced the results of the present study.

In children with CP, a decrease in EMG MDF of knee extensor, both RF and VL muscles, was found after several squat movements. This muscle fatigue also resulted in a more flexed posture in the initial squat position [30]. More recently, Eken et al. [7] reported muscle fatigue in children with CP at the end of a walking exercise. They highlighted muscle fatigue at the tibialis anterior, gastrocnemius medialis, and soleus muscles, but not at the quadriceps muscle as found in the present study [7]. This discrepancy, in terms of muscles affected by fatigue, could be explained by the difference in children gait pattern. Eken et al. [7] included a group walking with heterogeneous gait patterns [7], while the present study focused only on children walking in a crouch gait which requires important knee extensor work [5]. In a group of children with CP walking with various gait patterns, ankle power has been found to play a key role [31]. This ankle joint load may lead to calf muscle fatigue during a continuous walk, as reported by Eken et al. [7]. By focusing on a gait requiring excessive knee extensor work [5], the

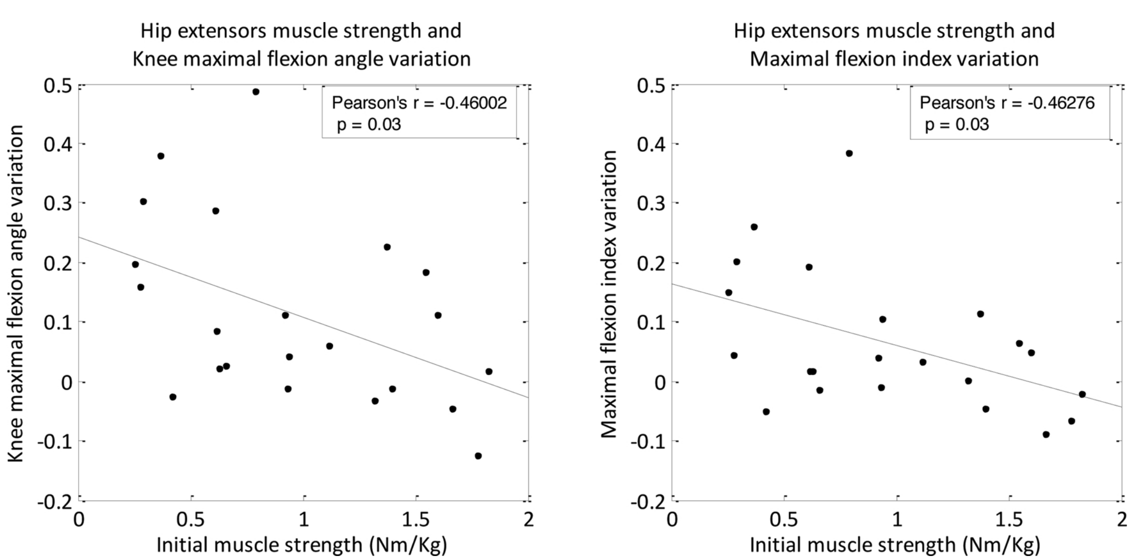

Fig. 3. Correlations between hip muscle strength and increased knee flexion and flexion index.

present study rather highlighted fatigue in knee extensor muscles. In addition to different gait patterns, the two groups of children had different GFMCS levels. Eken et al. [7] evaluated muscle fatigue in a group with mostly GMFCS level I (9/13 participants), whereas the present study included mostly children with GMFCS levels II and III (9/11 participants). Differences in gait parameters have been reported among GMFCS levels I to III [32]. Children with GMFCS level I exhibit more extended lower extremities during the stance phase of gait than those with GMFCS level III [32]. Hence, the muscle work required to walk varies between these two groups and may lead to differences in muscles affected by fatigue. Finally, during the 6mwe of the present study, gait was evaluated barefoot to avoid the influence of different types of shoes and orthosis on kinematic and EMG data. Eken et al. [7] evaluated EMG data when children were walking 5 min with their ankle-foot orthosis and regular shoes. This may have influenced the pattern of muscle activation [33] and thereby the MDF of EMG signals [34]. Based on their results, Eken et al. [7] recommended to focus on lower leg muscles in future research, since no fatigue was found in other muscles [7]. Considering the results of the present study, it would be relevant to continue to evaluate all muscles implicated in gait, as muscle fatigue may highly be influenced by gait pattern and severity of functional impairments. In our group of children with CP who walk in a crouch gait, muscle fatigue was found in RF muscle, when knee flexion was increased.

The increases in knee flexion and in flexion index were positively correlated with hip extensor weakness, but not with knee extensor group. These correlations suggest that strength in hip extensor could limit the increase in knee flexion at the end of a short continuous walk that results in RF muscle fatigue. This hypothesis is supported by a muscle-driven simulation study conducted by Steele et al. [24]. They identified that gluteus maximus is a primary contributor to hip extension [24] that indirectly facilitates knee extension in crouch gait. Furthermore, Arnold et al. [35] have suggested that gluteus maximus, among other muscles, play a key role in the acceleration toward knee extension during crouch gait [35]. Hence, weakness of this muscle group may result in an increase in knee flexion, as observed at the end of the 6mwe in the present study. For children with greater hip extensor strength, this muscle group may have contributed to limit the increase in lower limb flexion as muscle fatigue appeared in RF muscle. Strengthening hip extensor muscles in children who walk in a crouch gait might help to prevent the deterioration of the gait pattern during daily life activities.

The present study had some limitations to consider for interpretation. First, the study included a small sample of children with CP who walk in a crouch gait. However, the study included children with homogeneous gait, which reduces the number of children to include, but allows valuable results concerning this group. Second, muscle strength was measured during isometric muscle contractions, which is not task-specific to walking movement. It gives however an estimate of the capacity of the children to generate force at each joint. Finally, ankle muscle strength was not measured because the reliability of this measure is low with a handheld dynamometer method. Although no fatigue was found at ankle muscles, the relationship between plantarflexor strength and crouch gait adaptations at the end of a continuous walking exercise needs to be considered in future studies in order to complete the understanding of crouch gait adaptation during continuous walking.

In conclusion, an increase in crouch gait and RF fatigue were reported at the end of a 6-min walking exercise performed at comfortable speed in children with CP who walk in a crouch gait. The present study was the first to report RF muscle fatigue to explain this kinematic adaptation in crouch gait. The correlation between hip extensor strength and the kinematic changes suggest the key role of the hip extensor to limit lower limb flexion in children who walk in a crouch gait. Finally, continuous walking exercise is a relevant evaluation to consider in gait analysis to assess the adaptations which occur during daily locomotor activities.

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Conflict of interest statement

The authors had no conflict of interest.

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Appendix A. Supplementary data

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