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ORIGINAL PAPER

THE SAGITTAL PLANE KNEE PROPRIOCEPTION IN AMBULATORY INDIVIDUALS WITH CEREBRAL PALSY: A SINGLE-CENTER PILOT STUDY IN POLAND

CZUCIE POZYCJI STAWU KOLANOWEGO W PŁASZCZYŹNIE STRZAŁKOWEJ U CHODZĄCYCH PACJENTÓW Z MÓZGOWYM PORAŻENIEM DZIECIĘCYM: BADANIE PILOTAŻOWE

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ABSTRACT

Introduction

Proprioception is known to be affected in individuals with cerebral palsy (CP). However, it is still unclear how the static component of proprioception, joint position sense (JPS), is affected by the disease.

Aim

The aim of this study was to determine whether the knee JPS in the sagittal plane is impacted by CP.

Material and methods

Thirty-three participants were recruited for this study: 14 patients with spastic CP and 19 typically developing volunteers. JPS at 30˚ and 60˚ were tested and recorded by the electrogoniometer. The joint position error was defined as the error in the ability of a participant to replicate target lower limb position in the space.

Results

Mean joint position error was larger in the CP group for both joint angular positions. The statistically significant difference between the CP group and the control group was found only for 60˚. There is no significant difference between the mean values of joint position error at 30˚ and 60˚ in both CP and control groups.

Conclusion

CP affected joint position error of knee joint in the sagittal plane in ambulatory patients at 60˚ but not at 30˚. Our results suggested that the knee angular position does not affect the joint position sense error either in the CP or in typically developing individuals.

Keywords: proprioception, joint position sense, hypertonia, spasticity

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STRESZCZENIE

Wstęp

Zostało udowodnione, że propriocepcja może być zaburzona u osób z mózgowym porażeniem dziecięcym (MPD). Jednak nadal nie jest jasne, jak schorzenie to wpływa na statyczną składową propriocepcji – czucie pozycji w stawie.

Cel

Celem zaplanowanych badań było kreślenie wpływu MPD na czucie pozycji w stawie kolanowym, oraz określenie wpływu wartości kątowej jego ustawienia w płaszczyźnie strzałkowej na składową statyczną propriocepcji.

Materiał i metodyka

Do grupy badawczej zakwalifikowane zostały 33 osoby: 14 pacjentów z MPD oraz 19 zdrowych wolontariuszy – jako grupa kontrolna. Czucie pozycji w stawie kolanowym badane było przy pomocy elektrogoniometrów dla 30˚ i 60˚ zgięcia. Błąd pozycji stawu definiowany był jako błąd badanej osoby przy powtarzaniu zadanej pozycji stawu w przestrzeni.

Wyniki

Średni błąd pozycji stawu był większy w grupie osób z MPD dla obu badanych wartości kątowych. Statystycznie istotne różnice między grupą kontrolna a grupą pacjentów z MPD stwierdzone zostały jedynie dla wartości 60˚. Nie stwierdzono statystycznie istotnych różnic dla średniej wartości błędu pozycji w stawie dla 30˚ i 60˚ w obydwu grupach badawczych – grupie kontrolnej i grupie osób z MPD.

Wnioski

MPD upośledza czucie pozycji w stawie kolanowym w płaszczyźnie strzałkowej u chodzących pacjentów dla 60˚ i 30˚. Wartość kątowa pozycji w stawie kolanowym nie wpływa na wartość błędu pozycji w stawie w grupie pacjentów z MPD jak i w grupie kontrolnej.

Słowa kluczowe: propriocepcja, czucie pozycji w stawie, hipertonia, spastyczność

Introduction

Studies showed that proprioception is affected in individuals with cerebral palsy (CP) (Hohman *et al.*, 1958; Jones, 1976; Opila-Lehman *et al.*, 1985; Van Heest *et al.*, 1993; Wingert et al, 2009). The static component of proprioception is commonly tested by the joint position sense (JPS) test during the clinical physical examination which is used to evaluate individuals with different neurological disorders including CP (Goble, 2010). Deficits in JPS, with other common features of upper motor neuron lesions, which lead to structural impairments, deficits in motor control and thereby restricts the execution of functional tasks (Sanger et al, 2003; Sanger *et al.*, 2006). Recent studies of proprioception in CP were primarily focused on kinesthesia (i.e., the sense of limb movement), but only very few considered JPS (i.e., the static limb position). Moreover, previous studies were focused on upper limb JPS or rotational movement in the lower limb (Hohman *et al.*, 1958; Jones, 1976; Opila-Lehman *et al.*, 1985; Van Heest *et al.*, 1993; Wingert *et al.*, 2009). It is still unclear how JPS was affected in other joints and other anatomical planes.

The aim of this study was to determine whether the knee JPS in the sagittal plane is impacted by CP. A better understanding of JPS impact on knee movement in the sagittal plane range of motion during the gait (0°–60°) may provide insights to gait biomechanics and treatment strategies in CP individuals.

Materials and methods

The appropriate approval for this prospective study was acquired from the institutional review board. All subjects were tested in the institutional motion analysis laboratory. Written consents were obtained from all participants.

Participants

This study included two groups: individuals with a diagnosis of cerebral palsy (CP group), and typically developing individuals (control group). Participants in the CP group were ambulatory individuals who were classified as the Gross Motor Function Classification System (GMFCS) level I to III. To be enrolled in the study group, individuals need to meet the following inclusion criteria: (1) no trauma history; (2) no surgery or botulinum toxin injection within one year before the examination; and (3) able to understand and follow verbal instructions. The control group participants needed to meet the following inclusion criteria: (1) no neurological or physiological impairments, i.e., no damage in muscles, nerves, or ligaments; (2) no systemic disease or pain; (3) no trauma history; (4) within average physical activity level; and (5) not a high-performance athlete.

Protocols

Joint position sense (JPS): JPS for two angular positions was tested: 30° and 60° of knee flexion. During the JPS test, the subject was lying in the prone position on the examination table with both knees extended in the neutral position. Electrogoniometers (Biometrics Ltd., Newport, UK) were fixed to the lateral side (thigh and shank) of each lower limb to measure the sagittal plane knee joint angular position (i.e., flexion-extension). The electrogoniometer is a strain gauge which provides accurate angular values of the position between two body segments, which can avoid the inaccuracy in the traditional goniometer. Both sides were tested independently. The examiner flexed the knee of the right lower limb to 30° and hold it for 3 seconds, which was considered as the passive JPS (pJPS). The knee was passively extended to its neutral position. Next, the participant was instructed to actively repeat the previously demonstrated pJPS angle and hold it for 3 seconds; this knee flexion angle performed by the subject was considered as the active JPS (aJPS). Each subject underwent one training trial to ensure that he or she understood the instruction. The second trial was recorded and used in data analysis. The test for 60° knee flexion was performed afterward. Also, the left lower limb was tested with the procedure described above in both degrees. Data from both limbs were pooled together for the further data analysis.

Outcome measure: To determine the ability of a participant to replicate the target lower limb position demonstrated by the examiner without the visual assistance, a formula to calculate the joint position sense error (JPSe) was developed and used as the primary outcome measure in this study. The JPSe was defined as the percent error between aJPS and pJPS:

$$JPSe = \frac{|aJPS - pJPS|}{pJPS} \times 100\%$$

Data analysis

Analog signals from the electrogoniometer were digitized with the AxoScope software (Axon Instruments, Inc., Foster City, USA), using a 1084 Hz sampling rate. Further data analysis was performed with custom-written software based on MATLAB (MathWorks, Inc., Natick, USA). Kolmogorov–Smirnov test was used to verified that the data did not follow normal distribution. Therefore, a Kruskal–Wallis median test was used to establish an effect of disease on JPS at two knee flexion angular positions (30° and 60°). The Mann-Whitney test was used to compare JPSe between two groups, in each position.

The STATISTICA software (StatSoft, Inc., Tulsa, USA) was used for statistical analysis at the level of significance $p < 0.05$.

Results

In total, 33 subjects were recruited for this study: 14 individuals with spastic CP (9 females and 5 males) and 19 typically developing volunteers (14 females and 5 males). Anthropometric profiles (age, body mass, and height) of all subjects in the CP group and Control group were collected for all subjects (Table 1). The CP group consisted of 14 individuals with bilateral spastic involvement (age = > 14.6 ± 5.7 years; range: 7–28 years).

Table 1. Characteristics of study participants.

	Patient (n = > 14)	**Control** (n = > 19)
	Mean ± SD	Mean ± SD
Age (years)	14.6 ± 5.7	21.7 ± 1.5
Body Height (m)	1.5 ± 0.1	1.7 ± 0.1
Body Weight (kg)	41.4 ± 11.6	63.7 ± 9.3

SD = > Standard Deviation; n = > number of patients

The control group consisted of 19 typically developing volunteers (age = > 21.7 ± 1.6 years; range: 20–24 years). Data was collected on 27 limbs from the CP group and 38 limbs from typically developing subjects (Table 2). One limb from the CP group was excluded from the analysis because of incomplete data.

Table 2. Joint position sense error (JPSe) in the CP group and the control group.

	Patient (n = > 27)			**Control** (n = > 38)			*p*
	Mean ± SD	Min	Max	Mean ± SD	Min	Max	
JPSe (30°)	22.7 ± 18.4	1.1	84.5	13.4 ± 11.8	0.7	41.2	> 0.05
JPSe (60°)	20.6 ± 17.0	0.9	81.1	9.5 ± 6.5	0.0	27.4	**< 0.01**

n = > number of limbs; p = > level of statistical significance from the Kolmogorov–Smirnov test.

We found that the disease, CP, affects JPSe (Kruskal-Wallis test: H (1, N = > 62) = > 5.93, p = > 0.015). Mean JPSe was larger in CP group for both joint angular position: for 30˚ (22.7 ± 18.4 degree) and for 60˚ (20.6 ± 17.0 degree) than in control group (30˚: 13.4 ± 11.8; 60˚: 9.5 ± 6.5). The statistically significant difference between the CP group and the control group was found only at 60˚ ($p < 0.01$) (Table 2). Differences between the mean values of JPSe at 30˚ and 60˚ in both CP and control groups were not statistically significant (CP group: p = > 0.85, T = > 458, Z = > 0.18; control group: p = > 0.35, T = > 64, Z = > 0.96).

Discussion

The aim of this study was to determine whether the knee JPS in the sagittal plane is impacted by CP. Our data suggested that CP affects JPS. When mean values were compared, the JPSe in CP group was larger for both joint angular position than JPSe in typically developing subjects (for 30˚ up to 60%; for 60˚ up to 100%), but the difference was statistically significant only for 60˚. Moreover, we found that there was no statistically significant impact of knee angular position on JPSe in both CP and typically developing subjects (p = > 0.85 in the CP group; p = > 0.35 in the control group).

There was no previously reported data of knee JPS in the sagittal plane in CP individuals and how it was modulated by the angular position of the segments of the body. Moreover, the results presented in recent studies of JPS tested upper limb and other joints in the lower limb are unclear. There was no agreement in the estimation of the significance of the proprioception deficit (Hohman *et al.,* 1958; Jones, 1976). In general, recent data presented small to moderate decreases in JPS in CP individuals and suggested that deficits in proprioception may be a substantial problem in this population (Van Heest *et al.,* 1993; Wingert *et al.,* 2009). However, studies focused on the JPS deficit in CP patients showed that in 54–70% of the limbs, JPS was not affected, which was similar to the results described in healthy subjects (up to 60%) (Barrett *et al.,* 1991; Skatvedt, 1960; Van Heest *et al.,* 1993). It has been reported that in spastic CP individuals, proprioception was affected (10–15% reduced) but not as significantly as muscle strength (30% reduced) or tendon tap reflex reaction (over a dozen time increase) (Lebiedowska *et al.,* 2004; Wiley and Damiano, 1998; Wingert *et al.,* 2009). Our data also suggested that CP affected JPS (JPSe at 60˚ increase up to two folds) but may not be the crucial feature of motor function impairments in all CP individuals which were reported in previous studies (Lebiedowska *et al.,* 2004; Wiley and Damiano, 1998; Wingert *et al.,* 2009).

The previous study considered the factors which can modulate JPS (Wingert *et al.,* 2009). It was shown that vision, increase in the effort signal or dominant side of the body could impact the positional matching error. In our study, we tried to establish if the angular position of the joint modulates JPS and we found that the knee angular position in the sagittal plane does not affect JPS.

Moreover, the majority of previous studies treated CP subjects as a homogeneous group. Our analysis of JPS at 30˚ of knee flexion and monosynaptic reflexes showed that JPSe may increase if tendon tap reflexes are non-exaggerated and JPS of individuals with exaggerated monosynaptic reflexes is similar to JPS of typically developing subjects. It confirmed that the CP group is not homogenous and analysis of the means may not reflect the real deficits of JPS. Future studies should focus on more homogenous groups of individuals.

Limitations

There were some limitations in this present study. Our study focused on only one of the factors modulating the final movement possibilities of CP individuals and did not consider the influence of the most characteristic deficits in this group, e.g. exaggerated reflexes or spasticity, on JPSe. As a pilot study, the sample size is relatively small (CP group, n = > 14; control group, n = > 19). Because of the recruitment difficulty, demographics of participants between groups were not matched statistically. Moreover, our study included only ambulatory CP individuals with GMFCS level I to III, but not non-ambulatory individuals who are usually much more severely affected. Regarding the natural history of neurological maturation, the heterogenicity of wide age range in the CP group (7–28 years) may cause potential inconsistency of clinical presentations.

Future work should focus on gaining a better understanding of the origin of sensorimotor deficits not only describe their consequences. JPS is just one of many impairments (e.g., spasticity, hyperreflexia, hypertonia, and weakness) which leads to improper motor control in individuals with CP. Thus, potentially affected functional skills should be investigated. To better understanding the results, a research group should be as homogenous as possible in non-homogeneous diseases such as CP.

Conclusions

In summary, our data provided the argument that JPS of the knee joint is affected at 60˚ but not at 30˚ in the sagittal plane in ambulatory individuals with CP. Moreover, our results suggested that the knee angular position does not affect JPSe either in the CP or in typically developing individuals.

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