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Is the gait profile score a good marker of gait dysfunction in individuals with late effects of poliomyelitis?

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Abstract - Late effects of poliomyelitis (LEoP) are characterized by new gait abnormalities that occur many years after the initial poliomyelitis illness. Currently, there is no consensus on the most appropriate evaluation to detect gait disorders following LEoP. This study aimed to assess and compare the effectiveness of the gait profile score with that of the symmetry index (SI) to characterize gait abnormalities resulting from the LEoP. The SI for stance, swing, double-support duration and the step length, and gait profile score were computed from gait analysis of 12 poliomyelitis subjects and 12 healthy participants. Receiver operating characteristic analysis was used to measure the sensitivity and specificity of the SI and the gait profile score to discriminate patients with the LEoP and healthy participants. The area under the receiver operating characteristic curve was calculated for both gait the profile score and SI. With AUC values all above 0.83 (good discrimination), SI and GPS significantly discriminated the participants with the LEoP from the healthy participants (all p -values < 0.001). The results of this study show that both the gait profile score and SI may be used with a similar sensitivity by clinicians to identify potential gait abnormalities in patients with the LEoP.

Keywords: gait disorders, symmetry index, receiver operating characteristic

Résumé - Le Gait Profile Score est-il un bon indicateur des perturbations locomotrices chez le patient atteint de séquelles tardives de la poliomyélite? Les séquelles tardives de la poliomyélite (LEoP) sont caractérisées par l'apparition chez le patient de nouvelles perturbations locomotrices, survenant de nombreuses années après la primo-infection. Actuellement, il n'existe pas de consensus sur l'indice le plus approprié pour détecter et quantifier ces nouvelles perturbations. Cette étude vise donc à comparer la sensibilité et la spécificité de deux index d'évaluation locomotrice qui sont, l'indice de symétrie (IS) et le Gait Profile Score (GPS). Le GPS ainsi que l'IS de 4 paramètres locomoteurs (longueur de pas, % de phase d'appui, % de phase oscillante, % de phase de double support) ont été calculés à partir de l'analyse cinématique de 12 sujets post LEoP et de 12 sujets asymptomatiques. L'aire sous la courbe de la fonction d'efficacité du récepteur (courbe ROC en anglais) a été utilisée pour mesurer la sensibilité et la spécificité de l'IS et du GPS. Que ce soit pour l'IS des 4 paramètres locomoteurs ou le GPS les valeurs d'aire sous la courbe sont toutes supérieures à 0,83 (bonne discrimination). En d'autres termes, l'IS ou le GPS discriminent significativement les participants ayant des perturbations locomotrices post LEoP des participants asymptomatiques (toutes les valeurs $p < 0,001$). Les résultats de cette étude montrent que le GPS et l'IS peuvent être utilisés avec une sensibilité similaire par les cliniciens pour identifier les perturbations locomotrices des patients post LEoP.

Mots clés : perturbations locomotrices, indice de symétrie, courbe ROC

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1 Introduction

After many years, individuals with a history of poliomyelitis may develop new health problems. This phenomenon, called the “late effects of poliomyelitis” (LEoP), is characterized by the appearance of several symptoms including muscular fatigability, general fatigue, pain, muscle weakness and musculoskeletal pain during physical activity (Lexell & Brogårdh, 2012). These symptoms lead to several gait abnormalities (Genêt, *et al.*, 2010; Portnoy & Schwartz, 2013) that limit the mobility (Lexell & Brogårdh, 2012; Vreede, *et al.*, 2013).

The symmetry index (SI) is the most commonly clinical tool used to describe the degree of gait alteration in poliomyelitis (Arazpour, *et al.*, 2016; Portnoy & Schwartz, 2013), and it is associated with the risk of falling in this population (Portnoy & Schwartz, 2013). It involves comparison of the spatiotemporal parameters (*e.g.* stride length and swing phase duration) of the more affected side with that of the less affected side. However, patients with the LEoP may suffer from bilateral as well as unilateral after-effects (Schweizer, *et al.*, 2014). Even in the case of unilateral poliomyelitis, the contralateral side may be affected due to reduced mobility. Therefore, the SI may not be sufficient to describe gait abnormalities in patients with the LEoP. It is therefore critical to improve the clinical evaluation of gait in patients with the LEoP by establishing objective criteria providing the degree of their gait abnormalities in comparison with a healthy population. In this context, the use of the gait profile score (GPS), developed by Baker *et al.* (2009) and based on a single and global score including nine gait parameters (Baker, *et al.*, 2009), may be a better method of characterizing gait abnormalities in patients with the LEoP. Indeed, previous studies have demonstrated that some gait parameters used by the GPS are altered in patients with the LEoP. This is the case for the foot progression (Portnoy & Schwartz, 2013), the kinematic of the hip, knee, and ankle (Vreede, *et al.*, 2013). In addition, Schweizer *et al.* (2014) showed that the GPS is strongly associated with muscle strength for several pathologies, including poliomyelitis participants (Schweizer, *et al.*, 2014). The greater the muscle weakness, the higher the GPS.

The decision of a diagnostic test is often based on whether or not the value of a continuous variable exceeds a threshold value. In general, the individual is referred to as ‘sick’ or ‘not sick’. It is possible that the diagnostic test will give a positive result for a not sick person or a negative result for a sick person. In this context, sensitivity and specificity are two basic measures of diagnostic accuracy. Sensitivity is the proportion of true positives that are correctly identified by the test. Specificity is the proportion of true negatives that are correctly identified by the test (Pepe, 2003). Among the different metrological approaches that assess the quality of a diagnostic test, the receiver operating characteristic (ROC) analysis is a tool that simply describes the range of compromises obtained by a diagnostic test. The ROC analysis is widely used in biomedical studies to assess the diagnostic accuracy of a continuous variable (Ma, Bandos, & Gur, 2015).

To recommend the best management options for these patients (*e.g.* gait devices, surgery or rehabilitation), it appears critical to know which clinical tools are the most discriminant to assess their gait abnormalities. As a result of specific muscle weakness in patients with the LEoP (Schweizer, *et al.*, 2014), we hypothesized that the GPS would be more discriminant to characterize gait abnormalities in patients with the LEoP than the SI. For this purpose, we compared the ability of the SI and the GPS to discriminate patients with the LEoP from healthy individuals.

2 Method

Data was obtained from twelve patients with the LEoP (7 men and 5 women; age: 53 ± 15 years, height: 172 ± 11 cm, body mass: 67 ± 14 kg; mean \pm SD) who were seen in the Motion & Gait Analysis Lab at APH-HP Raymond Poincaré teaching hospital Garches in France from February 2012 through to June 2017. All patients had to free from gait assistance devices. Twelve healthy subjects (7 men and 5 women; age: 31 ± 7 years, height: 171 ± 6 cm, body mass: 64 ± 7 kg) were retrospectively included in this study, that was in conformity with the Declaration of Helsinki (last modified in 2013).

Participants with the LEoP were asked to walk barefoot on a 10 m walkway at their spontaneous speed. Gait was analyzed using a motion-capture system (Motion Analysis Corporation, Santa Rosa, CA, USA; sampling frequency: 100 Hz). The trajectories of 24 markers placed on anatomical landmarks using the Helen Hayes model were collected and filtered using a fourth-order zero-lag Butterworth low-pass filter (6 Hz cut-off frequency) (Collins, *et al.*, 2009). For each participant, spatiotemporal and joint kinematic parameters were extracted using Orthotrak 6.2.8 (Motion Analysis Corporation, Santa Rosa, CA, USA) using a minimum of 10 gait cycles.

The SI for stride length, percent of stance phase, percent of swing phase, and percent of double support were calculated according to the equation proposed by Kim & Eng (2003). The SI is the difference between the more affected side and the less affected side for patients with the LEoP and is the difference between the non-dominant side and the dominant side for the healthy participants. For both groups, all SIs were calculated from an average step, including a minimum of 10 gait cycles. The GPS was obtained from nine kinematic parameters: pelvic tilt, pelvic obliquity, pelvic rotation, hip flexion, hip rotation, knee flexion, ankle dorsiflexion and the foot progression angle (Baker, *et al.*, 2009). It corresponds to the root mean square of these nine kinematic parameters between the patients with the LEoP and the healthy participants, according to Baker *et al.* (2009). For the patients with the LEoP the GPS was computed for the most affected leg, and for the healthy group, it was computed for the dominant leg.

Data and statistical analyses were performed using Matlab (R14, The MathWorks Inc., Natick, USA). The Shapiro-Wilk test revealed that the GPS and the SI were

Table 1. Gait disorder scores for patients with late effects of poliomyelitis and for healthy participants. Legend: LEOp group = group including patients with late effects of poliomyelitis; SI = symmetry index; GPS = gait profile score. The GPS is expressed in degree. For the LEOp group, the GPS is computed for the most affected leg, and for the healthy group, it is computed for the dominant leg. The SI is the difference between the more affected side and the less affected side for patients with the LEOp and is the difference between the non-dominant side and the dominant side for the healthy participants. All results are presented as mean \pm standard deviation.

| | LEOp group ($n = 12$) | Healthy group ($n = 12$) | p -value |
|-----------------------|----------------------------|-------------------------------|-------------|
| SI stride length (cm) | 18.28 \pm 25.18 | 2.90 \pm 2.50 | $p = 0.002$ |
| SI stance phase (%) | 11.78 \pm 9.02 | 1.98 \pm 1.31 | $p = 0.003$ |
| SI swing phase (%) | 20.44 \pm 16.60 | 3.00 \pm 1.97 | $p = 0.002$ |
| SI double support (%) | 15.26 \pm 10.14 | 5.17 \pm 4.78 | $p = 0.006$ |
| GPS ($^{\circ}$) | 8.36 \pm 3.65 | 5.13 \pm 0.77 | $p = 0.006$ |

not normally distributed. Non-parametric Mann-Whitney U tests were used for the GPS and the SI to compare patients with the LEOp and healthy participants. The ROC analysis was used to measure the sensitivity and specificity of the SI and the GPS to discriminate patients with the LEOp and healthy participants. The Youden index is used to plot the ROC curve of a classifier. The true positive rate (sensitivity) is plotted as a function of the false positive rate ($1 - \text{Specificity}$) for cut-off points in a ROC curve (Landais, Besson, & Jais, 1994). The area under the curve (AUC) describes the test's overall performance. A value of 1 indicates perfect discrimination, a value > 0.9 is considered as excellent discrimination, a value between 0.7 and 0.9 is considered as good discrimination and a value of 0.5 indicates poor discrimination. To compare the ability of the SI and the GPS to discriminate participants with the LEOp from healthy subjects, we used the approach proposed by DeLong, DeLong & Clarke-Pearson (1988). This method calculates the covariance matrix of the AUCs, and then the variance of the difference between the two AUCs and the associated p -value based on a normality assumption are calculated. For all statistical tests, the significance level was set at $p < 0.05$.

3 Results

The SI and the GPS were significantly ($p < 0.001$) higher in the LEOp group than in the healthy participant group (Tab. 1).

Figure 1 shows the ROC curves for the SI and the GPS. With AUC values all above 0.83 (good discrimination), SI stride length, SI stance phase, SI swing phase, SI double support and GPS significantly discriminated the participants with the LEOp from the healthy participants (all p -values < 0.001 ; Tab. 2). Statistical analysis revealed no significant difference in the AUC values between all the SI parameters and the GPS (all p -values > 0.61) (see Fig. 1).

4 Discussion

This study showed that the SI and the GPS allow discriminating gait disorders in patients with the LEOp

from healthy individuals. However, contrary to our expectations, the GPS is not superior to the SI in discriminating between these populations.

To the best of our knowledge, the GPS of patients with the LEOp was presented for the first time in the current study. We found higher GPSs in the LEOp group than in the healthy group. Since the GPS includes several spatiotemporal and kinematic parameters of gait, this result showed that overall gait is altered in patients with the LEOp. In addition, the ROC analysis showed that the GPS is able to discriminate (AUC = 0.83) patients with the LEOp from healthy individuals. Therefore, the GPS appears to be a good measure to assess gait alterations in patients with the LEOp.

While gait was nearly symmetrical in the healthy group, it was clearly asymmetrical for patients with the LEOp, with SIs varying from 12% (stance phase) to 20% (swing phase). These SIs for patients with the LEOp are close to those reported in previous studies using the same method (Arazpour, *et al.*, 2016; Portnoy & Schwartz, 2013). In addition, the ROC analysis showed that the SIs for stride length, stance phase, swing phase and double support presented AUC values > 0.83 , indicating that all the SIs successfully discriminated patients with the LEOp from healthy individuals. Our results therefore confirmed that the gait profile of individuals is appreciably characterized by asymmetries.

The statistical analysis failed to demonstrate any differences between the SIs of gait parameters and the GPS to discriminate patients with the LEOp and healthy subjects. Despite statistical differences for the AUC values, the GPS tends to be the least discriminatory measure with a sensitivity of only 0.67. Similar to the study of Morel *et al.* (2017) who reported that the GPS may not identify gait abnormalities in low-disability multiple sclerosis patients (Morel, *et al.*, 2017), we assumed that the greater between-participant variability in the LEOp group may explain, at least in part, the low sensitivity of the GPS. This could be explained by the fact that the GPS is based on the median value of the gait parameters rather than on the variability of these parameters (Morel, *et al.*, 2017). A limitation of this is the age difference between the two groups. Regarding SI, it

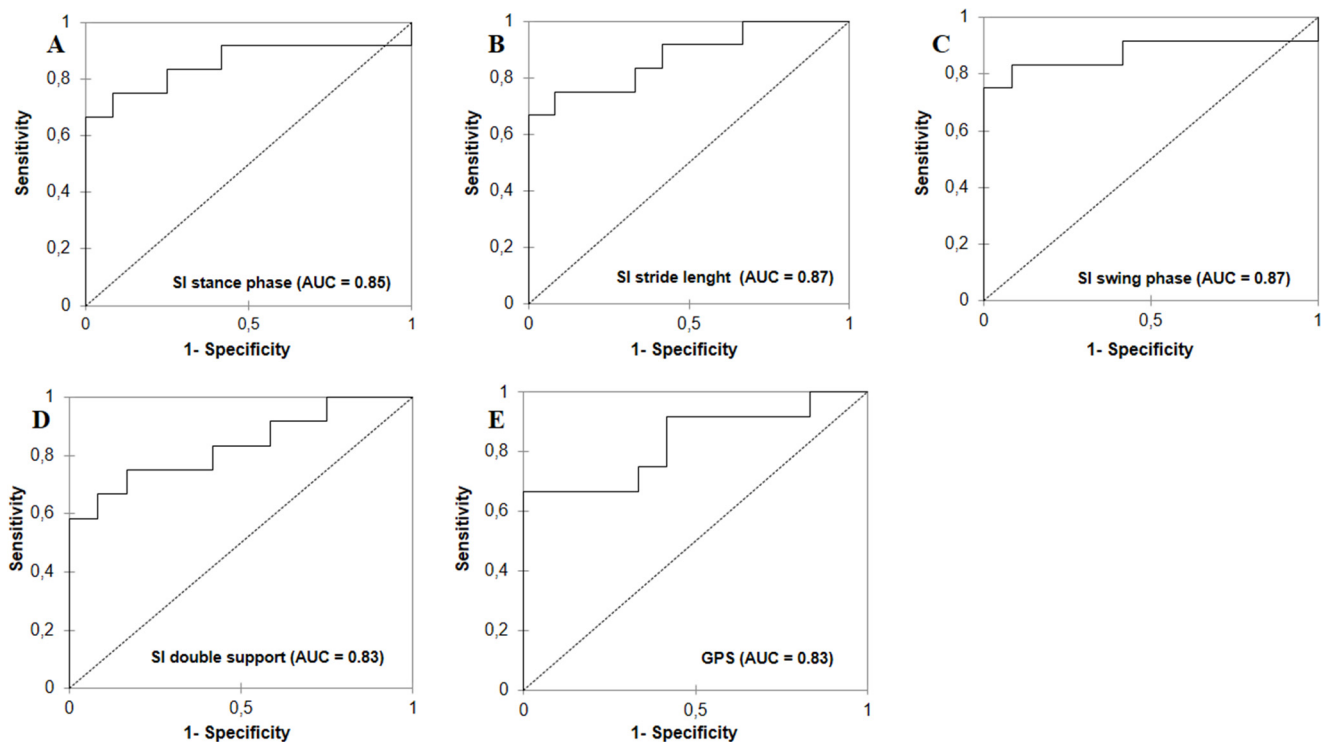


Fig. 1. Receiver operating characteristic (ROC) curves for all gait disorder scores. Legend: (A) ROC curve for SI of the stance phase. (B) ROC curve for SI of the stride length. (C) ROC curve for SI of the swing phase. (D) ROC curve for SI of double support. (E) ROC curve for the GPS. SI = symmetry index; GPS = gait profile score; AUC = area under curve; SI = symmetry index.

Table 2. Receiver operator characteristic curve (ROC) analysis for symmetry indexes and gait profile score. Legend: ROC = receiver operating curve; AUC = area under curve; CI = confidence interval; SI = symmetry index; GPS = gait profile score; PPV = positive predictive value; NPV = negative predictive value.

| | AUC (95% CI) | Optimal criterion | Sensitivity (95% CI) | Specificity (95% CI) | PPV | NPV | <i>p</i> -value |
|-----------------------|-----------------|----------------------|-------------------------|-------------------------|------|------|-----------------|
| SI stride length (cm) | 0.87 | 5.69 | 0.75 | 0.92 | 0.90 | 0.79 | $p < 0.001$ |
| SI stance phase (%) | 0.85 | 3.64 | 0.75 | 0.92 | 0.90 | 0.70 | $p < 0.001$ |
| SI swing phase (%) | 0.87 | 4.99 | 0.83 | 0.92 | 0.91 | 0.85 | $p < 0.001$ |
| SI double support (%) | 0.83 | 9.81 | 0.75 | 0.83 | 0.82 | 0.77 | $p < 0.001$ |
| GPS (°) | 0.83 | 6.30 | 0.67 | 1.00 | 1.00 | 0.75 | $p < 0.001$ |

has already been found that healthy aging does not alter the SI at spontaneous walking speed (Malone & Bastian, 2016; Patterson, et al., 2012). This supports that the SI difference we observed between healthy group and LEOp group cannot be related to the age difference between the two groups. The effect of aging on GPS has not yet been investigated. In a recent work, it has been reported a GPS of $5.93^\circ \pm 1.16^\circ$ in a sample of healthy people aged 48.9 ± 15.0 years (Coghe, et al., 2020). Despite an about 18-year age difference, this GPS is close to the one we observed in our healthy group (5.13 ± 0.77). On the contrary, it is substantially smaller to the GPS we observed in our LEOp group (8.36 ± 3.65) with an age difference of only 4 years. This supports that the GPS difference we observed between healthy group and LEOp group was mainly due to the

specificity of each groups (LEOp *vs.* healthy status), even though age difference could be slightly involved in this GPS difference.

In conclusion, the results of this study show that both the GPS and SIs may be used with a similar sensitivity by clinicians to identify potential gait abnormalities in patients with the LEOp. The SI comparing spatiotemporal parameters of gait and the GPS including several kinematic parameters of gait, these tools may be used in a complementary manner to provide to the clinician a more complete picture of gait abnormalities for each patient with the LEOp. To strengthen their validity, it remains to determine the sensitivity to change of these clinical assessment tools under varying potential clinical management (*e.g.* gait assistance devices, muscle strengthening, pharmacological treatments).

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Author contribution statement

Anthony Supiot: conception and design of study Data collection, drafting the article, final approval of the version to be published. François Genêt: conception and design of study, data analysis and interpretation, final approval of the version to be published. Thomas Cattagni: drafting the article, critical revision of the article, final approval of the version to be published. Marjorie Salga: critical revision of the article, final approval of the version to be published. Nicolas Roche: critical revision of the article, final approval of the version to be published. Didier Pradon conception and design of study data collection, final approval of the version to be published.

Conflict of interest

The authors declare that they have no conflicts of interest in relation to this article.

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