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RESEARCH PAPER

Daily activities, participation, satisfaction, and functional mobility of adults with cerebral palsy more than 25 years after selective dorsal rhizotomy: a long-term follow-up during adulthood

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ABSTRACT

Purpose: To determine changes in level of accomplishment and satisfaction in daily activities and social participation, and functional mobility in adults with cerebral palsy (CP) more than 25 years after selective dorsal rhizotomy (SDR).

Materials and methods: This long-term observational nine-year follow-up study included 26 adults (median age 35 years) with CP and spastic diplegia, and 26 matched typically developing adults. Assessment tools used were the Life-Habits questionnaire and the Functional Mobility Scale.

Results: Most of the adults with CP were independent and satisfied with accomplishing life habits and no changes were determined, except for a small change in the Housing accomplishment level. Compared to typically developing adults, the CP cohort was more dependent in accomplishing Mobility and Recreation. However, the level of satisfaction was similar for most life habits except for Mobility. Functional mobility did not change, but correlated with Life-Habits results.

Conclusions: Adults with CP showed high and stable levels of accomplishment and satisfaction in daily activities and social participation more than 25 years after SDR. This is in contrast with the literature, where functional decline was shown for individuals with CP as they age. The relation with functional mobility highlights the importance to focus the rehabilitation on maintaining walking ability in order to enable high level of daily activities and social participation in adults with CP.

➤ IMPLICATIONS FOR REHABILITATION

- Selective dorsal rhizotomy (SDR) is a valuable treatment option for a selective group of children with cerebral palsy (CP) in order to reduce spasticity.
- The long-term outcomes of SDR on level of accomplishment and satisfaction in daily activities and social participation as well functional mobility in adults with CP are not clear.
- More than 25 years after SDR adults with CP experienced stable and lasting high levels of functioning regarding daily activities and social participation and were satisfied with the way they accomplished life habits.
- Functional mobility was correlated to level of accomplishment and satisfaction in daily activities and social participation, which highlights the importance to focus rehabilitation programs on maintaining functional mobility in order to enable daily activities and social participation in adults with CP.

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Cerebral palsy; rhizotomy; activities of daily living; social participation; mobility limitation; ageing

Introduction

Until recently, the primary focus of cerebral palsy (CP) treatment and research has focused on children and less on adults. As life expectancy of individuals with CP has become similar to that of typically developing adults, adults with CP are now considered one of the world's largest populations with a physical impairment [1]. Therefore, healthy ageing with CP and the effects of interventions received in childhood have become a important topic to monitor.

CP management has shifted, according to the current International Classification of Function, Disability and Health model [2], to emphasize a biopsychosocial approach. This means that in addition to the treatment of "body structure and function" impairments, the level of "activity and participation" is acknowledged as an important factor within the lifelong management plan for people with CP [3].

One of the biggest challenges with healthy ageing is to prevent or minimize the secondary effects of CP on the musculoskeletal system and to improve functional status and quality of life throughout one's life span (e.g., prevent contractures, bone deformities, and pain) [4]. Spasticity is one of the main contributors to the

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development of secondary complications and is estimated to be present in approximately 80% of people with CP [5].

An effective treatment regime to reduce spasticity is a neurosurgical procedure known as selective dorsal rhizotomy (SDR) [4,6,7]. SDR reduces spasticity by transecting a percentage of lumbar rootlets which disrupts the reflex arc at spinal cord level. First described by Foester in 1913, SDR only gained popularity after Professor Warrick Peacock re-introduced an adapted technique of this procedure in Cape Town, South Africa in the 1980s and in the USA from 1986 [8]. If strict selection criteria are adhered to, SDR has shown to be an effective treatment to reduce spasticity and improve functionality [9]. This has found to have a positive effect on the child's participation in the community [4,10]; however, less is known about the activity, participation and satisfaction levels of adults with CP who underwent SDR in childhood.

A study by Langerak et al. [11], interviewed a group of adults with CP more than 17 years after SDR, reported that most of these adults were independent in accomplishing daily activities and social participation with high satisfaction levels. In line with this, Munger et al. [12] reported similar levels of participation in their SDR (more than 10 years post-SDR) and control group (adults with CP who did not undergo SDR) based on the Frequency of Participation Questionnaire. A study by van der Slot et al. [13], reported slightly poorer outcomes with perceptions of a low health-related quality of life in a group of adults with CP and spastic diplegia (though not indicating what treatment was received in childhood). In addition, Benner et al. [14], who also did not report on performed childhood interventions, observed increased health concerns (functional deterioration, pain, and severe fatigue) and an impact of these health concerns on activities in adults with CP. Studies reporting on the level of accomplishment and satisfaction in daily activities and social participation in adults with CP in developing countries are limited and longitudinal studies documenting on this subject more than 25 years following SDR do not exist.

Therefore, the aim of the study was to conduct a long-term follow-up study (2008–2017) to determine changes in level of accomplishment and satisfaction in daily activities and social participation and functional mobility, in a group of adults with CP and spastic diplegia who underwent SDR more than 25 years ago. A secondary aim was to compare these outcomes of adults with CP to a matched group of typically developing adults. Third, to determine associations between the subjects' current level of accomplishment and satisfaction in daily activities and social participation with: (i) subjects' characteristics and (ii) level of functional mobility.

Methods

Study design and subjects

This observational nine-year follow-up of adults with CP who underwent SDR during childhood more than 25 years ago. The baseline study was performed in 2008 [11] and was based on 32 adults with CP, who underwent SDR at Red Cross War Memorial Children's Hospital in Cape Town, South Africa, between 1981 and 1991.

The subjects were selected for SDR based on strict selection criteria as described by Peacock and Arens [15,16]. They were diagnosed with spastic diplegia, without dystonia, athetosis, ataxia, and/or hypotonia. The aim of SDR was to improve on functional level, and therefore can be classified as Gross Motor Function Classification System (GMFCS) level I, II, or III pre-operatively [17]. They had to have sufficient trunk and lower extremity muscle strength, access to ongoing physiotherapy before and after SDR and adequate care-taker support.

Subjects from this 2008 study were contacted and asked if they were willing to participate in the current study (2017). In addition, a group of typically developing adults was recruited and matched for gender, age, body mass index (BMI), and socio-economic status (SES). Adults in the typically developing group were not included if they had any neuromuscular disorders and/or other physical impairments. Before enrolling into the study, all subjects signed a written informed consent. The study was approved by the Human Research Ethics Committee of the University of Cape Town.

All the assessments (interview, measurements, observations, and questionnaire) were conducted by one of the two investigators (BEV and NGL) using strict guidelines. NGL was the principal investigator of the 2008 study, and familiarized BEV with the outcome measures, using the same guidelines as in baseline study. If a clear judgment could not be made by the one investigator, the other investigator was consulted.

Subject characteristics

Subjects' socio-demographic information and indicators of participation in daily life were obtained by a semi-structured interview. Age, gender, SES, marital status, children, living situation, highest level of education attained, employment status, main source of income, and current health status were captured as part of this interview. SES was estimated based on housing density, as suggested by Micklesfield et al. [18], which is calculated by dividing the "number of people living in the house" by the "number of rooms in the house" (excluding kitchen and bathroom). Score categories are as follows <1: "high SES"; ≥ 1 and ≤ 1.5: "normal SES"; and >1.5: "low SES" [18]. In addition, to the interview, subjects' BMI (based on measured height and weight) and level of GMFCS [17] (based on observation and consultation with the subject) were determined.

Life habits questionnaire

The Life Habits (LIFE-H 3.1) questionnaire was used to evaluate subjects' level of accomplishment in daily activities and social participation, as well as how satisfied they were to accomplish these life habits [19]. The LIFE-H questionnaire consists of 77 life habits, which are divided into 12 subscales related to Daily Activities and Social Roles. Daily Activities includes the following six subscales: "Nutrition", "Fitness", "Personal care", "Communication", "Housing", and "Mobility", while Social Roles includes the subscales of "Responsibilities", "Interpersonal relationships", "Community life", "Education", "Employment", and "Recreation". For each of the Daily Activities and Social Roles subscales, a weighted Accomplishment and Satisfaction score was calculated [19,20].

The accomplishment scores range from 0 (not accomplished or achieved) to 9 (accomplished without difficulty and without assistance) and are based on the degree of difficulty and the type of assistance required to accomplish a task, while the satisfaction scores range from "very unsatisfied" (score −10) to "very satisfied" (score 10) [20]. The LIFE-H questionnaire has shown to be reliable and valid [19,20] and has been used in different cohorts studies in adults with CP [11,13,21,22]. In line with previous study of Langerak et al. [11], the subscale of "Education" was excluded from the analyses.

Functional Mobility Scale

The Functional Mobility Scale (FMS) was used to determine subjects' level of mobility in their daily environment (performance level). The FMS is based on a six-level ordinal grading system, rating the mobility for three different distances, namely 5, 50, and 500 m, while taking the use of an assistive device in consideration. The FMS scores range from using a wheelchair (score 1) to being totally independent on all surfaces (score 6) [23]. The FMS has been used before in adults with CP [24] and has shown to be a valid and reliable scale to determine functional mobility [23,25].

Statistical analysis

Data were analyzed using SPSS 25 (SPSS Inc., Chicago, IL) [26]. Data were assessed for normality using the Shapiro–Wilk test of normality. Since data were not normally distributed, non-parametric two-tailed inferential tests were used. Statistical analyses were performed by PPV, who is affiliated with the Methodology & Statistics section of the Department of Psychology and Child Studies at Erasmus University Rotterdam and also performed the statistical analyses for the 2008 study [11]. Descriptive statistical analyses were used to summarize subjects' characteristics and indicators of participation (see Table 1).

Life-H data were categorized for interpretation purposes. Similar as in the 2008 study, the weighted accomplishment scores of the LIFE-H questionnaire were divided into three categories: (I) score $\geq$8.0: independent with no difficulties (with or without assistance); (II) score 5–8: independent with difficulties (with or without assistance); (III) score $\leq$5.0: dependent, as this life habit is not performed by the subject or carried out with human assistance. The results of the weighted satisfaction scores were categorized into two levels: (I) score $<$0.0: dissatisfied and (II) score $\geq$0.0: satisfied.

For statistical analyses, the numerative (non-categorized) scores were used and for the descriptive statistics medians and interquartile ranges (IQRs) were reported. Wilcoxon's rank tests were used to compare the 2008 and 2017 CP cohorts on the subscales and total scores of accomplishment and satisfaction levels. To determine differences between the current CP and typically developing cohort, Mann–Whitney's *U* tests were used. As a threshold for statistical significance, to control for multiple comparisons (48), a Bonferroni corrected alpha-level of $p \leq 0.001$ was applied for both analyses. Furthermore, for statistically significant results, 95% CI of the median differences were reported.

In line with the 2008 study, FMS scores were categorized for interpretation purposes, to: (I) independent/able to walk without walking aids (FMS levels 5 and 6); (II) need to use walking aids (FMS levels 2, 3, and 4); and (III) wheelchair dependent (FMS level 1). For statistical analyses, the non-categorized scores were used. Wilcoxon's rank tests were applied to compare the 2008 and 2017 CP cohorts on the FMS scores. As a threshold for statistical significance, to compensate for multiple comparisons (three), a Bonferroni corrected alpha-level of $p \leq 0.0167$ was applied. Furthermore, for statistically significant results, 95% CI of the median differences were reported.

Spearman's rank correlation analyses were used to examine the associations between LIFE-H scores (i.e., total accomplishment and satisfaction scores), subject characteristics values (age at SDR, current age, SES, BMI) and FMS scores of the adults with CP. As a threshold for statistical significance, to control for multiple comparisons (eight and six), a Bonferroni corrected alpha-level of respectively $p \leq 0.006$ and $p \leq 0.008$ was applied.

Results

Subjects' characteristics

From the 32 subjects who participated in the 2008 study [11], six subjects were not included in the 2017 study. One subject was pregnant, one had been injured in a motor vehicle accident, three choose not to participate and one was lost to follow-up. The characteristics of the remaining 26 participants are shown in Table 1. In addition, similar information is also provided for the adults with CP at the 2008 study and the 26 typically developing adults.

At the nine-year follow-up (period ranging from 25 to 35 years after SDR), GMFCS levels of adults with CP were stable, except for one subject who improved one level (from GMFCS level III to GMFCS level II). The adults with CP in the current study and typically developing adults were comparable in age, gender, SES, and BMI. The SES within both cohorts was similar, with 8% (CP) and 11% (typically developing) being classified as having a low SES, 42% (CP) and 35% (typically developing) a normal SES and 50% (CP) and 54% (typically developing) a high SES. Both cohorts also showed a comparable distribution of subjects' BMI with 3.8% (CP) and 0% (typically developing) underweight, 50% (CP) and 42% (typically developing) normal BMI, and 50% (CP) and 58% (typically developing) overweight.

The majority of the CP study cohort had no other diagnosis influencing their medical status. Health issues reported were hypertension ($n = 4$), Crohn's disease ($n = 1$), Graves' disease ($n = 1$), asthma ($n = 1$), and mental health conditions (e.g., depression, anxiety) ($n = 3$). Incontinence was reported in three subjects, all of whom had urge incontinence; a male subject had incontinence preceding SDR which remained unchanged, and two females reported incontinence following pregnancy and delivery.

Table 1. Subjects' characteristics of CP ($n = 26$) and typically developing cohorts ($n = 26$).

Variable	2008 CP *n* (%)/median [IQR]	2017 CP *n* (%)/median [IQR]	2017 typically developing *n* (%)/median [IQR]
Gender, male	16 (60)	16 (60)	16 (60)
Age (years)	26.8 [25.4–32.5]	35.8 [34.2–41.4]	35.7 [33.2–44.2]
SES	1.25 [0.8–1.7]	0.9 [0.7–1.3]	0.8 [0.6–1.3]
BMI (kg/m^2)	23.0 [20.3–29.9]	25.2 [21.6–31.2]	25.9 [24.1–28.2]
Age at SDR (years)	4.9 [3.7–10.1]	4.9 [3.7–10.1]	
Follow-up time (years)	21.4 [18.4–23.8]	30.1 [27.5–32.7]	
GMFCS			
Level I	13 (50)	13 (50)	
Level II	9 (35)	10 (38)	
Level III	4 (15)	3 (12)	

IQR: interquartile range; SDR: selective dorsal rhizotomy; SES: socio-economic status; BMI: body mass index; GMFCS: Gross Motor Function Classification System.

Indicators of participation

The indicators of participation of the adults with CP (2008 and 2017 study) and the typically developing adults are shown in Table 2. With ageing, more adults with CP moved out the house of (grand)parents (35% versus 62%), were in a relationship or married (46% versus 65%) and had children (19% versus 42%). This number of adults with CP being in a relationship was similar to typically developing adults (61%), while it was more common for typically developing adults (of similar age) to be a parent (77%) and not living with (grand)parents (89%). Also more typically developing adults were employed (89%) compared to the adults with CP in both studies (69% and 73%).

Life habits questionnaire

The accomplishment and satisfaction levels, based on the LIFE-H questionnaire, of each cohort are shown in Figure 1(A,B), respectively. Overall adults with CP in the current study were independent and satisfied with accomplishing daily activities and social roles. However, 19% of the adults with CP scored being dependent for "Mobility" activities, while 12% scored to be dependent for "Community Life", 15% for "Employment", and 12% for "Recreation". In line with this, 8% of the subjects with CP were dissatisfied with their "Mobility", 4% with their "Community Life", 12% with their "Employment" status, and 4% with their "Recreation" activities.

No differences in the Total Accomplishments and Satisfaction level scores were found between the 2008 and the current study in the adults with CP. However, when analyzing the different subscales between the 2008 and the 2017 study, adults with CP were more dependent in accomplishing "Housing" activities ($p < 0.001$) (Table 3). The median difference was −1.1 (95% confidence interval [CI]: −1.56 to −0.76). Within the existing score range of 10, this difference in accomplishment of "Housing" activities can be considered as small. Furthermore, the width of the 95% CI is small indicating a rather precise parameter estimate.

In comparison to adults with CP, the typically developing adults had overall higher accomplishment levels for daily activities and social roles ($p < 0.001$). The median difference of accomplishment levels for daily activities was 0.8 (CI: −0.5 to 3.9) and for social roles was 0.6 (CI: −0.8 to 3.1). Within the existing score range of 10, this difference in accomplishment of daily activities and social roles can be considered as small. However, the width of the 95% CI is large indicating a rather limited precision of the parameter estimate. Significant differences ($p < 0.001$) were also found in several subscales, with median differences of 0.6 (CI: −2.2 to 4.2) for "Fitness", 0.1 (CI: 0.0–3.7) for "Personal care", 1.3 (CI: −1.3 to 5.6) for "Housing", 2.9 (CI: −0.4 to 6.7) for "Mobility" and 2.3 (CI: −0.6 to 7.9) for "Recreation". Within the existing score range, the median differences in the accomplishment of "Fitness" and "Personal Care" can be considered as small while the differences in accomplishment of "Housing", "Mobility", and "Recreation" between the CP and typically developing group can be considered as medium and more clinically relevant. However, the width of the 95% CI is large indicating a rather limited precision of the parameter estimate. In contrast to accomplishment, satisfaction levels were the same between the adults with CP and the typically developing adults, except for the "Mobility" subscale levels ($p < 0.001$) (Table 3). The median difference was 4.0 (CI: −3.3 to 13.8). Within the existing score range of 20, for satisfaction, this difference in satisfaction with "Mobility" can be considered as a medium. However, the width of the 95% CI is large indicating a rather limited precision of the parameter estimate.

Table 2. Subjects' indicators of participation for CP and typically developing cohorts.

Variable	2008 CP *n* (%)	2017 CP *n* (%)	2017 typically developing *n* (%)
Marital status			
Single	14 (54)	9 (35)	9 (35)
Divorced/widow	0 (0)	0 (0)	1 (4)
Relationship	7 (27)	5 (19)	2 (7)
Married	5 (19)	12 (46)	14 (54)
Children			
0	21 (81)	15 (58)	6 (23)
1	3 (11)	6 (23)	8 (31)
≥2	2 (8)	5 (19)	12 (46)
Living situation			
Living on own	2 (8)	2 (8)	7 (27)
With (grand) parents	17 (65)	10 (38)	3 (11)
With partners	6 (23)	14 (54)	15 (58)
With others (e.g., family, friends)	1 (4)	0 (0)	1 (4)
Educational attainment			
Primary	1 (4)	1 (4)	0 (0)
Secondary	9 (35)	7 (27)	6 (23)
Higher education	16 (61)	18 (69)	20 (77)
Employment			
Paid employed	17 (65)	17 (65)	21 (81)
Self employed	1 (4)	2 (8)	2 (8)
Unemployment (health reason)	5 (19)	0 (0)	0 (0)
Unemployment (other reason)	2 (8)	7 (27)	3 (11)
Student	1(4)	0 (0)	0 (0)
Main income			
Paid job	19 (73)	17 (65)	23 (89)
Disability grant	6 (23)	9 (35)	0 (0)
Family income and other	1 (4)	0 (0)	3 (11)

Functional Mobility Scale

Walking performance over 5, 50, and 500 m determined by the FMS in adults with CP at the 2008 and 2017 study are shown in Figure 2. No changes in the FMS scores were found between the 2008 study and the current study.

Within the current study, 65% of the subjects was able to walk independently (FMS score 5 or 6) over 5 m with 54% being able to walk independently over 50 and 500 m. Walking aids were used (FMS score 2, 3, or 4) by 35% of the subjects over a 5-m distance, while 46% used it over a 50-m distance and 42% needed a walking aid for a 500 m walking distance. One adult with CP (4%) needed a wheelchair (FMS score 1) to cover 500 m at the moment of the current study.

Associations

The estimated rank correlation coefficients between the LIFE-H questionnaire, accomplishment and satisfactions levels, and subject characteristics are shown in Table 4. No relationships between subject characteristics and the level of accomplishment and satisfaction were found.

The estimated rank correlation coefficients between the LIFE-H questionnaire and FMS scores are shown in Table 5. Strong correlations were found between accomplishment levels and FMS scores, with correlations ranging from 0.85 to 0.89. In line with this, strong correlations were also found between the level of satisfaction and FMS scores, with correlations ranging from 0.73 to 0.79.

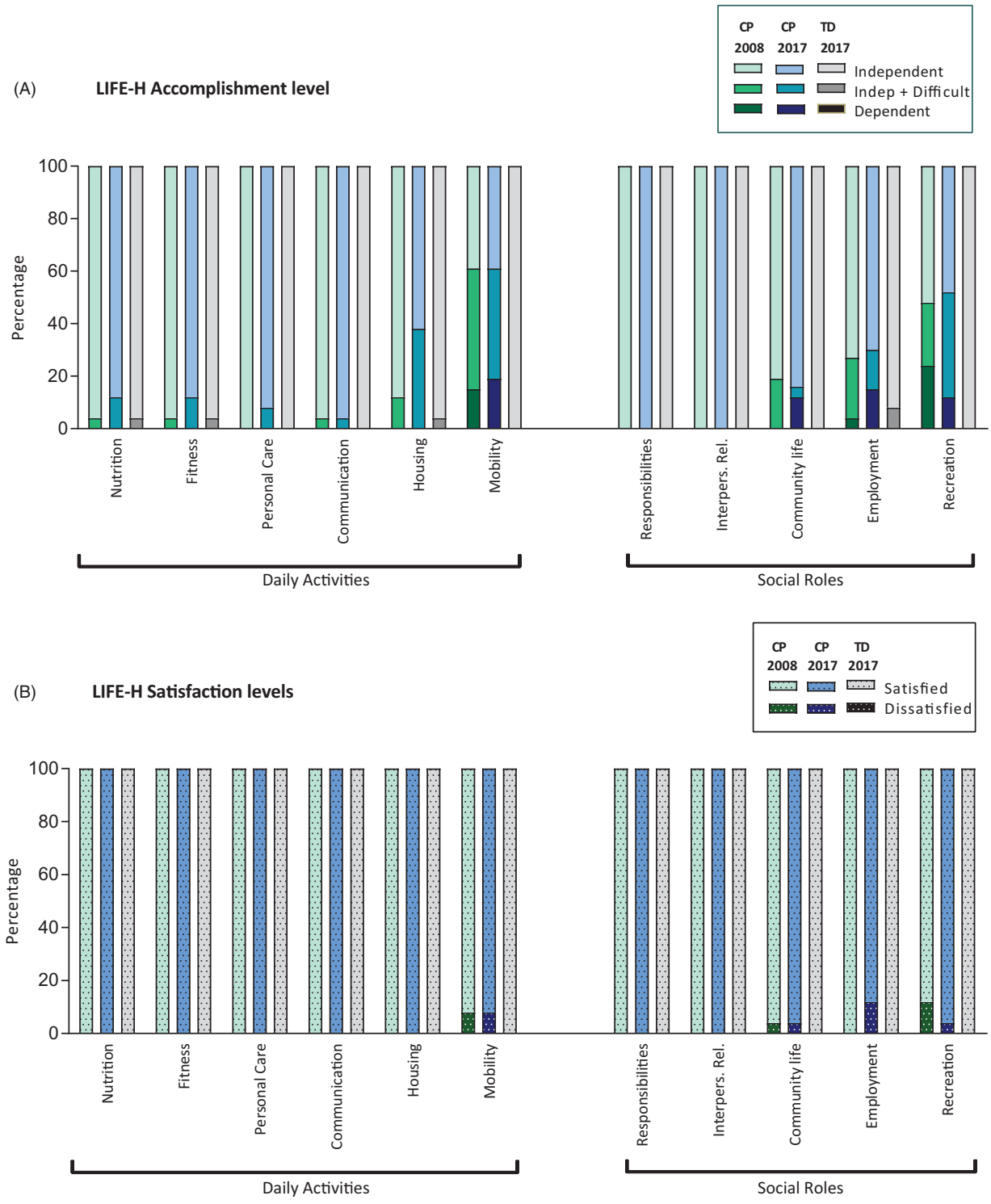

Figure 1. Outcomes of LIFE-H categorized for CP (2008 and 2017) and typically developing cohorts. (A) Accomplishment levels weighted score ≥8.0: independent and no difficulties; score 5–8: independent with difficulties; and score ≤5.0: dependent or not able to accomplish. (B) Satisfaction levels with score ≥0.0: satisfied; and score <0.0: dissatisfied.

Discussion

With the knowledge of secondary problems occurring with age in CP [1], there is an importance to understand the long-term outcomes of interventions performed in childhood like SDR. This is the first long-term follow-up study reporting the level of accomplishment and satisfaction in daily activities and social participation as well as functional mobility for adults with CP and spastic diplegia more than 25 years after SDR.

Level of accomplishment and satisfaction in daily activities and social participation

More than 25 years after SDR, most of the adults with CP were independent and satisfied with their accomplishment of daily activities and participation in the community.

No change was found between the 2008 [11] and the 2017 study regarding the overall level of accomplishment and satisfaction in daily activities and social participation. For the

Table 3. Outcomes of the LIFE-H questionnaire for the CP and typically developing cohorts.

	Accomplishment			Satisfaction		
LIFE-H item	2008 CP Median [IQR]	2017 CP Median [IQR]	2017 typically developing Median [IQR]	2008 CP Median [IQR]	2017 CP Median [IQR]	2017 typically developing Median [IQR]
Daily activities						
Nutrition	10.0 [10.0–10.0]	10.0 [9.7–10.0]	10.0 [10.0–10.0]	10.0 [8.4–10.00]	10.0 [8.8–10.00]	10.0 [8.8–10.0]
Fitness	9.6 [7.5–10.0]	9.4 [7.5–10.0]#	10.0 [10.0–10.0]#	7.5 [5.0–10.0]	7.5 [5.4–9.4]	9.4 [5.0–10.0]
Personal care	10.0 [9.7–10.0]	9.9 [9.5–10.0]#	10.0 [10.0–10.0]#	10.0 [6.4–10.0]	9.3 [6.8–10.0]	10.0 [9.8–10.0]
Communication	10.0 [9.7–10.0]	10.0 [10.0–10.0]	10.0 [9.9–10.0]	10.0 [8.6–10.0]	10.0 [10.0–10.0]	10.0 [10.0–10.0]
Housing	9.8 [9.3–10.0]*	8.3 [7.3–9.8]*,#	10.0 [8.9–10.0]#	10.0 [5.8–10.0]	9.1 [5.0–10.0]	10.0 [9.0–10.0]
Mobility	6.8 [5.5–9.0]	7.0 [5.7–9.3]#	10.0 [10.0–10.0]#	6.1 [3.3–8.0]	6.0 [1.2–8.5]#	10.0 [8.6–10.0]#
Total daily activities	9.2 [8.9–9.7]	9.0 [8.4–9.6]#	9.9 [9.7–10.0]#	8.7 [5.8–9.5]	8.1 [6.1–9.4]	9.5 [8.6–10.0]
Social roles						
Responsibilities	10.0 [10.0–10.0]	10.0 [10.0–10.0]	10.0 [10.0–10.0]	10.0 [9.3–10.0]	10.0 [9.8–10.0]	10.0 [9.8–10.0]
Interpersonal relationships	10.0 [10.0–10.0]	10.0 [10.0–10.0]	10.0 [10.0–10.0]	10.0 [8.3–10.0]	10.0 [7.9–10.0]	10.0 [8.4– 10.0]
Community life	10.0 [8.5–10.0]	10.0 [8.9–10.0]	10.0 [10.0–10.0]	8.0 [5.0–10.0]	10.0 [5.5–10.0]	10.0 [9.7– 10.0]
Employment	9.0 [7.7–10.0]	9.3 [7.2–10.0]	10.0 [9.9–10.0]	7.1 [5.0–10.0]	7.2 [5.0–10.0]	10.0 [8.8– 10.0]
Recreation	8.6 [5.2–10.0]	7.4 [6.4–10.0]#	10.0 [10.0–10.0]#	6.4 [4.3–8.6]	7.1 [4.8–10.0]	10.0 [8.5–10.0]
Total social roles	9.1 [8.1–9.8]	8.9 [8.1–9.6]#	10.0 [9.8–10.0]#	7.8 [6.3–9.6]	8.5 [5.5–9.5]	9.7 [8.5–10.0]

IQR: interquartile range.
As a threshold for statistical significance a Bonferroni corrected alpha-level of $p \leq 0.001$ was applied for all scores.
*Significant difference between 2008 and 2017 CP.
#Significant difference 2017 CP and 2017 typically developing adults.

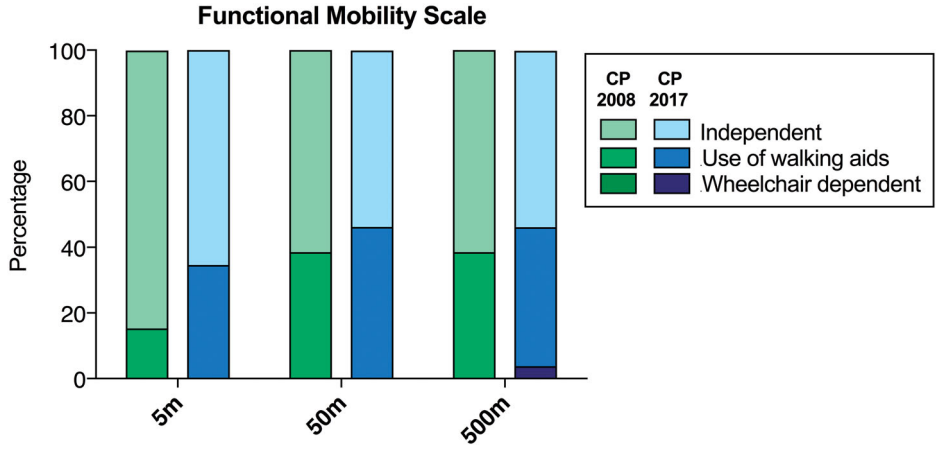

Figure 2. Functional mobility scores for CP cohorts. Categorized FMS scores: (I) independent/able to walk without walking aids (FMS levels 5 and 6); (II) need to use walking aids (FMS levels 2, 3, and 4); and (III) wheelchair dependent (FMS level).

Table 4. Correlations (Spearman's rho) between LIFE-H and subjects' characteristic variables for 2017 CP cohort.

	Age at SDR		Current age		SES		BMI	
LIFE-H total scores	*r*	*p*	*r*	*p*	*r*	*p*	*r*	*p*
Accomplishment	−0.040	0.846	−0.225	0.270	−0.309	0.124	−0.171	0.405
Satisfaction	−0.086	0.678	−0.236	0.246	−0.175	0.391	−0.114	0.580

SDR: selective dorsal rhizotomy; SES: socio-economic status; BMI: body mass index.
As a threshold for statistical significance, a Bonferroni corrected alpha-level of $p \leq 0.006$ was applied.

Table 5. Correlations (Spearman's rho) between LIFE-H and FMS scores for 2017 CP cohort.

	FMS – 5 m		FMS – 50 m		FMS – 500 m	
LIFE-H total scores	*r*	*p*	*r*	*p*	*r*	*p*
Accomplishment	0.85	<0.001	0.89	<0.001	0.87	<0.001
Satisfaction	0.79	<0.001	0.73	<0.001	0.76	<0.001

FMS: Functional Mobility Scale.
As a threshold for statistical significance, a Bonferroni corrected alpha-level of $p \leq 0.008$ was applied.

subdomains, the only change was found in the Housing subscale (accomplishment), though the difference was relatively small. The change in this score over years was mainly attributed to a higher percentage of "major household tasks" and "maintaining the grounds of their home" were performed by a proxy. This may be explained by the fact that more adults in the 2017 study were married and living with their partners instead of their (grand)parents. Another reason could be, with the increase of SES, that subjects were possibly able to pay a third party to complete these tasks done. Interestingly, none of the subjects were dissatisfied with the level of accomplishment for this subscale, which suggests that they were happy to have or pay someone else to complete the household task.

The perceived stability in levels of accomplishment in daily activities and social participation in the current study is in contrast with the literature, where functional decline was shown for individuals with CP as they age [27,28,29]. The mean age at which functional deterioration has been reported is approximately 37 years [30]. On average, our current cohort was younger (median: 35 years), though the study was based on a nine-year follow-up and the age at the 2017 study ranged from 33 to 53 years. The stability of level of accomplishment and satisfaction

in daily activities and social participation during ageing was also confirmed by the statistical analyses reporting no correlation between the Total Life-H accomplishment and satisfaction scores and age. A reason for absence of functional deterioration in our CP cohort, might be that all subjects underwent an SDR (where strict selection criteria have been adhered). The SDR may assist in avoiding the secondary complications associated with spasticity reduction, and has been found to have a positive effect on level of functioning [10].

Overall adults with CP showed some differences for daily activities and social roles subscales compared to their typically developing peers. These differences were relatively small; however, the differences for accomplishment of Mobility and Recreation were more substantial showing that adults with CP were more dependent and faced more difficulties for these domains compared to the typically developing adults. This can be explained by the fact that 50% of our subjects with CP were classified as GMFCS level II or III. However, the reported 61% of the adults with CP that experienced difficulties with Mobility in our study is lower than the 77% determined in a study of van der Slot et al. [13], who studied adults with spastic diplegia aged 25–45 years. This in contrast to Donkervoort et al. [22], who reported percentages of 27%, but their study cohort was much younger (16–20 years) and also included hemiplegia (50%) and quadriplegia (25%). Both studies did not report on interventions given in childhood.

Another interesting element in this study was the difference in percentage of adults with CP involved in competitive employment between this study (developing country) and other studies in adults with CP living in Western societies (developed countries). In the 2017 study, 73% of the CP cohort were employed, while in developed countries this ranged between 29 and 54% [13,31–33]. This was also reflected in the LIFE-H outcomes (subscale Employment) with 70% of the adults living in Cape Town, South Africa, being independent (score <8) and 56% of the adults with CP living in the Netherlands [13]. This disparity appears to be due to differences in social security systems and the value of disability grants between developing and developed countries.

Remarkably, despite these obstacles, adults with CP were as satisfied as their typically developing peers for all subscales (except for Mobility). This is relevant information for clinicians, realizing that adults with CP who are restricted in some activities and/or participation in daily life, might be satisfied with how they accomplish their life-habits. However, this does not apply for the Mobility domain. Adults with CP reported feeling restricted and less satisfied than TD peers regarding their mobility, suggesting that improving level of mobility should be emphasized at follow-up and/or rehabilitation programs.

Functional mobility

The majority (54%) of the adults with CP in the current study were able to walk independently without walking aids. Andersson and Mattsson [27] and Ando and Ueda [28] reported similarly results with 49% and 57%, respectively, where interventions in childhood were not specified. However, Maanum et al. [34] reported 70% of the study cohort being able to walk independently, although their study included also younger subjects (age range: 18–65 years) and individuals diagnosed with unilateral CP (almost 50% of their study population).

No change was found in FMS scores of the adults with CP between the 2008 and 2017 study. This is remarkable since most literature reports functional walking decline in subjects with CP while ageing, starting in their 20s and 30s [34]. A correlation between functional mobility (FMS score) and the level of accomplishment and satisfaction in daily activities and social participation was found. The fact that there was no change in FMS levels of adults with CP in the 2008 and the 2017 study, might be a contributing factor to the fact that there was also no significant change in level of accomplishment and satisfaction in daily activities and social participation. The current study showed that FMS performance can explain 72–80% of variance in overall accomplishment levels, and 54–63% of the variance in overall satisfaction levels. These findings highlight the importance for adults with CP to maintain their walking function and mobility in order to enable daily activities and social participation. Clinicians and therapists should therefore inform adults with CP about the importance of an active lifestyle.

Limiting factors

The sample size of the current study was moderate. However, as a first follow-up study in adults with CP more than quarter of a century after SDR, the conclusion should be seen as an important indication of long-term outcomes of SDR. The current study was conducted in South Africa, while results of similar studies from other research groups were based on study populations from developed countries. We should be aware that the health services in South Africa are not as well established as in Western countries [35]. To limit the influence of this factor, a reference cohort consisting of South African typically developing adults was included in this study. However, for comparison purposes, it would be interesting for future studies to include a reference cohort of South-African adults with CP and spastic diplegia, who did not receive SDR, but only orthopedic interventions or conservative treatment for example. The last point to address is that this study only focused on associations with a few personal characteristics (e.g., age, SES, BMI), while insight into other factors associated with level of accomplishment and satisfaction in daily activities and social participation (e.g., pain, fitness levels, depression) are important to guide future therapies.

In conclusion, this nine-year follow-up study in adults with CP and spastic diplegia reports positive outcomes regarding level of accomplishment and satisfaction in daily activities and social participation as well as functional mobility more than 25 years after SDR. Adults with CP and spastic diplegia have stable and lasting high levels of functioning within the Activity and Participation components of the International Classification of Function, Disability and Health model. In addition, they are overall satisfied with accomplishing daily activities and social participation in the community. This is relevant information for parents, care-givers and clinicians when they consider SDR as a treatment option for a child with CP. In addition, the strong correlation between functional mobility and the LIFE-H scores highlights the importance of focusing therapies and rehabilitation in adults with CP on maintaining their functional mobility in order to enable daily activities and social participation.

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References

[1] Haak P, Lenski M, Hidecker MJ, et al. Cerebral palsy and aging. Dev Med Child Neurol. 2009;51(Suppl. 4): 16–23.
[2] World Health Organization. International classification of functioning, disability and health: ICF. World Health Organization; 2001.
[3] Colver A, Fairhurst C, Pharoah PO. Cerebral palsy. Lancet. 2014;383(9924):1240–1249.
[4] Narayanan UG. Management of children with ambulatory cerebral palsy: an evidence-based review. J Pediatr Orthoped. 2012;32(Suppl. 2):S172–S181.
[5] Blair E, Watson L. Epidemiology of cerebral palsy. Semin Fetal Neonat Med. 2006;11(2):117–125.
[6] Novak I, McIntyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. Dev Med Child Neurol. 2013;55(10):885–910.
[7] Aquilina K, Graham D, Wimalasundera N. Selective dorsal rhizotomy: an old treatment re-emerging. Arch Dis Child. 2015;100(8):798–802.
[8] Langerak NG, Lamberts RP, Fieggen AG, et al. Selective dorsal rhizotomy: long-term experience from Cape Town. Childs Nerv Syst. 2007;23(9):1003–1006.
[9] Langerak NG, Lamberts RP, Fieggen AG, et al. Functional status of patients with cerebral palsy according to the International Classification of Functioning, Disability and Health model: a 20-year follow-up study after selective dorsal rhizotomy. Arch Phys Med Rehabil. 2009;90(6):994–1003.
[10] Health Quality Ontario. Lumbosacral dorsal rhizotomy for spastic cerebral palsy: a health technology assessment. Ont Health Technol Assess Ser. 2017;17(10):1–186.
[11] Langerak NG, Hillier SL, Verkoeijen PP, et al. Level of activity and participation in adults with spastic diplegia 17–26 years after selective dorsal rhizotomy. J Rehabil Med. 2011; 43(4):330–337.
[12] Munger ME, Aldahondo N, Krach LE, et al. Long-term outcomes after selective dorsal rhizotomy: a retrospective matched cohort study. Dev Med Child Neurol. 2017;59(11): 1196–1203.
[13] van der Slot WM, Nieuwenhuijsen C, van den Berg-Emons RJ, et al. Participation and health-related quality of life in adults with spastic bilateral cerebral palsy and the role of self-efficacy. J Rehabil Med. 2010;42(6):528–535.
[14] Benner JL, Hilberink SR, Veenis T, et al. Long-term deterioration of perceived health and functioning in adults with cerebral palsy. Arch Phys Med Rehabil. 2017;98(11): 2196–2205.
[15] Arens LJ, Peacock WJ, Peter J. Selective posterior rhizotomy: a long-term follow-up study. Child's Nerv Syst. 1989; 5(3):148–152.
[16] Peacock WJ, Arens LJ, Berman B. Cerebral palsy spasticity. Selective posterior rhizotomy. Pediatr Neurosurg. 1987; 13(2):61–66.
[17] Palisano RJ, Rosenbaum P, Bartlett D, et al. Content validity of the expanded and revised gross motor function classification system. Dev Med Child Neurol. 2008;50(10):744–750.
[18] Micklesfield LK, Levitt NS, Carstens MT, et al. Early life and current determinants of bone in South African children of mixed ancestral origin. Ann Hum Biol. 2007;34(6):647–655.
[19] Fougeyrollas P, Noreau L, Bergeron H, et al. Social consequences of long term impairments and disabilities: conceptual approach and assessment of handicap. Int J Rehabil Res. 1998;21(2):127–141.
[20] Noreau L, Lepage C, Boissiere L, et al. Measuring participation in children with disabilities using the assessment of life habits. Dev Med Child Neurol. 2007;49(9):666–671.
[21] Donkervoort M, Wiegerink DJ, van Meeteren J, et al. Transition to adulthood: validation of the Rotterdam Transition Profile for young adults with cerebral palsy and normal intelligence. Dev Med Child Neurol. 2009;51(1): 53–62.
[22] Donkervoort M, Roebroeck M, Wiegerink D, et al. Determinants of functioning of adolescents and young adults with cerebral palsy. Disabil Rehabil. 2007;29(6): 453–463.
[23] Graham HK, Harvey A, Rodda J, et al. The functional mobility scale (FMS). J Pediatr Orthoped. 2004;24(5):514–520.
[24] Lennon N, Church C, Miller F. Patient-reported mobility function and engagement in young adults with cerebral palsy: a cross-sectional sample. J Child Orthop. 2018;12(2): 197–203.
[25] Harvey A, Baker R, Morris ME, et al. Does parent report measure performance? A study of the construct validity of the Functional Mobility Scale. Dev Med Child Neurol. 2010; 52(2):181–185.
[26] Corporation I. SPSS statistics 25. Armonk (NY): IBM Corporation; 2017.
[27] Andersson C, Mattsson E. Adults with cerebral palsy: a survey describing problems, needs, and resources, with special emphasis on locomotion. Dev Med Child Neurol. 2001; 43(02):76–82.
[28] Ando N, Ueda S. Functional deterioration in adults with cerebral palsy. Clin Rehabil. 2000;14(3):300–306.
[29] Andren E, Grimby G. Dependence in daily activities and life satisfaction in adult subjects with cerebral palsy or spina bifida: a follow-up study. Disabil Rehabil. 2004;26(9): 528–536.
[30] Opheim A, Jahnsen R, Olsson E, et al. Walking function, pain, and fatigue in adults with cerebral palsy: a 7-year follow-up study. Dev Med Child Neurol. 2009;51(5): 381–388.
[31] Benner JL, Hilberink SR, Veenis T, et al. Course of employment in adults with cerebral palsy over a 14-year period. Dev Med Child Neurol. 2017;59(7):762–768.

[32] Michelsen SI, Uldall P, Kejs AM, et al. Education and employment prospects in cerebral palsy. Dev Med Child Neurol. 2005;47(8):511–517.

[33] Murphy KP, Molnar GE, Lankasky K. Employment and social issues in adults with cerebral palsy. Arch Phys Med Rehabil. 2000;81(6):807–811.

[34] Maanum G, Jahnsen R, Froslie KF, et al. Walking ability and predictors of performance on the 6-minute walk test in adults with spastic cerebral palsy. Dev Med Child Neurol. 2010;52(6):e126–e132.

[35] Donald KA, Kakooza AM, Wammanda RD, et al. Pediatric cerebral palsy in Africa: where are we? J Child Neurol. 2015;30(8):963–971.