# The power of Para sport: the effect of performance-focused swimming training on motor function in adolescents with cerebral palsy and high support needs (GMFCS IV) – a single-case experimental design with 30-month follow-up

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## ABSTRACT

**Objective** This study aims to evaluate the effect of a performance-focused swimming programme on motor function in previously untrained adolescents with cerebral palsy and high support needs (CPHSN) and to determine whether the motor decline typical of adolescents with CPHSN occurred in these swimmers.

**Methods** A Multiple-Baseline, Single-Case Experimental Design (MB-SCED) study comprising five phases and a 30-month follow-up was conducted. Participants were two males and one female, all aged 15 years, untrained and with CPHSN. The intervention was a 46-month swimming training programme, focused exclusively on improving performance. Outcomes were swim performance (velocity); training load (rating of perceived exertion min/week; swim distance/week) and Gross Motor Function Measure-66-Item Set (GMFM-66). MB-SCED data were analysed using interrupted time-series simulation analysis. Motor function over 46 months was modelled (generalised additive model) using GMFM-66 scores and compared with a model of predicted motor decline.

**Results** Improvements in GMFM-66 scores in response to training were significant ($p<0.001$), and two periods of training withdrawal each resulted in significant motor decline ($p\leq0.001$). Participant motor function remained above baseline levels for the study duration, and, importantly, participants did not experience the motor decline typical of other adolescents with CPHSN. Weekly training volumes were also commensurate with WHO recommended physical activity levels.

**Conclusions** Results suggest that adolescents with CPHSN who meet physical activity guidelines through participation in competitive swimming may prevent motor decline. However, this population is clinically complex, and in order to permit safe, effective participation in competitive sport, priority should be placed on the development of programmes delivered by skilled multiprofessional teams.

**Trial registration number** ACTRN12616000326493.

### WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Compared with ambulant people with cerebral palsy, gross motor function declines in non-ambulant people with cerebral palsy and high support needs (CPHSN). These patients are also less physically active, and it is plausible that relative inactivity contributes to motor decline; however, this premise has not been investigated.

### WHAT THIS STUDY ADDS

⇒ This study demonstrated that previously inactive adolescents with CPHSN who undertook performance-focused swimming training with multiprofessional guidance over 46 months improved sports performance and maintained gross motor function during a life stage when population-based modelling predicted gross motor decline.

### HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ This study introduces the novel concept of 'Para Sport as Medicine' and suggests that performance-focused sports training programmes delivered by multiprofessional teams may be an effective means of preventing motor decline among people with CPHSN, as well as conferring a range of psychosocial and well-being benefits.

## BACKGROUND

Cerebral palsy (CP) is the most common neuromotor disorder affecting children and non-progression of the underlying neuropathology is a defining feature of CP.[1] In children with CP who are ambulant—Gross Motor Function Classification System (GMFCS) levels I and II—gross motor function improves from birth to approximately 7–9 years of age and then plateaus. However, among children with CP who are non-ambulant, have high support needs and are classified as GMFCS levels IV and V (CPHSN), early developmental gains are generally followed by a decline in motor function throughout adolescence and into early adulthood.[2] The underlying causes of this decline are poorly understood although it has been suggested that reduced access to neurological care, the development of new neurological conditions[3] or poor management of hypertonia during periods of growth[4] may contribute.

We posit that insufficient habitual physical activity may contribute to motor function decline in adolescents with CPHSN. Specifically, the majority of children and adolescents with CP are insufficiently active for good health[5] and, compared with those who are ambulant, those with CPHSN are more sedentary and less physically active.[6] It is plausible that the relatively greater gross motor decline of adolescents with CPHSN is caused, at least in part, by their relatively low levels of habitual physical activity.

Unfortunately, people with CPHSN are grossly underrepresented in exercise training studies. A recent review identified that only 3% of participants were either GMFCS level IV or V.[7] While evidence indicates that physical activity can improve gross motor function in people with CP, the effect has only been demonstrated in children at GMFCS levels I and II,[8] and as they do not experience the motor decline associated with GMFCS IV and V, the generalisability of the finding is not known. Additionally, interventions have been brief (8–12 weeks) with limited follow-up, so the extent to which improvements are maintained is not known.[8]

Competitive Para swimming is a type of physical activity open to people with CPHSN, and it provides them with critical avenues to engage in competitive swimming, one of Australia's most popular and culturally significant sports, particularly among children and adolescents.[9] Para swimmers devote the majority of their sports participation time to performance-focused training, defined as training that is planned and undertaken for the primary purpose of maximising sports performance.[9] Because of its focus on performance enhancement—primarily maximising swimming velocity[10]—performance-focused swimming training is clearly distinct from other conventional aquatic therapies (including hydrotherapy) which explicitly focus on therapeutic outcomes and which have been shown to be effective.[11] However, while performance-focused training does not have therapeutic goals, personal testimony from experienced Para swimmers with CP indicates they attribute large, meaningful improvements in physical function to such training.[9] However, to date, the veracity of this testimony has not been evaluated in swimmers with CP, including swimmers with CPHSN.

Investigating whether performance-focused swimming training prevents motor decline in people with CPHSN presents considerable challenges. Severe functional limitations increase the time cost of participation for people with CPHSN by 8–13 times,[12] significantly increasing research costs. Further, the heterogeneity that characterises CP is greatest in this population, who are often affected by a greater number of comorbidities that are more severe[13] and many of these comorbidities (eg, seizure disorders, eating and drinking difficulties and pain) act as independent prognostic variables in exercise training trials. In group-based research designs such as randomised controlled trials (RCTs), the result of this heterogeneity is predictable, systematic between-participant differences in exercise training responses which act to amplify noise and threaten internal validity.

Single-case experimental research designs (SCEDs), where each participant acts as their own control, account for logistical and research design challenges in people with CPHSN and offer a methodologically robust alternative to RCTs.[14] The SCED generates high-level evidence (equivalent to RCTs[15]) using small samples, permits tailoring of the intervention to each participant and produces individual outcomes—the SCED is one of the few designs in which it is possible to detect if, when and to what extent each participant responds to the intervention.[16] These design features are particularly advantageous for studies in people with CPHSN: a relatively small, heterogeneous population who require tailored interventions and support which meets their personal needs, and who have been largely excluded from the literature to date.[10]

Therefore, this study employed a single-case experimental design to address two primary aims: to evaluate the effect of a performance-focused swimming programme on gross motor functioning in previously untrained, inactive adolescents with CPHSN and to determine whether the motor decline typical of adolescents with CPHSN occurred in swimmers who trained and competed regularly for one Paralympic cycle over a 46-month period.

## METHODS

The ParaSTART (Sports Training And Research Team) programme was established to facilitate research presented in this manuscript and other projects. 'Para' indicates a focus on people who are eligible to compete in Para sport.[17] The programme specialises in physically demanding Para sports training for people with high support needs—those using wheeled mobility and requiring personal assistance for fundamental tasks of daily living. A brief vignette is available here—https://youtu.be/HxCRf7hHj7k.

### Participants

Young, inactive people with CPHSN were recruited from a 30 km radius from the University of Queensland, Brisbane, Australia. Key inclusion and exclusion criteria are described fully in the published protocol.[10] Participants were one female and two males with CPHSN, aged 15–16 years on enrolment, classified as GMFCS IV. None were achieving WHO physical activity guidelines, and they had not previously participated in performance-focused sports training. Two other participants were screened and did not meet inclusion criteria, and one other participant was screened and excluded due to contraindication to the intervention (see online supplemental appendix 1). Included participants provided assent, and participants' parents/guardians provided informed consent on enrolment. Table 1 describes clinical characteristics, sport classes and stroke preference of each participant.

### Study design

To evaluate the effect of a performance-focused swimming programme on gross motor functioning, a Multiple-Baseline, Single-Case Experimental Design (MB-SCED) was used. It took place over 16 months, between March 2017 and July 2018, and comprised five phases A1 (baseline)-B1-A2-B2-A3: where 'A' phases represent periods of no training or training withdrawal, and 'B' phases represent training exposures, each being 16 weeks duration, all standard training block duration for competitive swimmers.[10 18] Two features of this design make it particularly strong. First, there are repeated measures throughout all phases—a total of 102 data collection points, exceeding the 75 data points required for this design according to SCED guidelines.[19] Second, the transitions between training and withdrawal phases were temporally staggered,[10] and the 5-phase design presented a total of 12 opportunities to detect an experimental effect, 4 transitions for each participant (from an A-B or B-A phase). The MB-SCED methods are reported fully in the published protocol.[10] The trial was registered (Australian and New Zealand Clinical Trial Registry number ACTRN12616000326493).

Following the MB-SCED, a 30-month follow-up period commenced during which participants continued a schedule of regular training and monitoring. Data from the full 46 months

**Table 1** Participant characteristics

| Participant | Age | | Sex | Neurological subtype/ distribution | Function (GMFCS*; MACS†; CFCS‡; VFCS§; EDACS¶ | Comorbidities**/medical events | Preferred stroke | Para swimming class |
|---|---|---|---|---|---|---|---|---|
| | At enrolment | At study cessation | | | | | | |
| 1 | 15 years, 2 months | 19 years, 0 months | Male | Spastic Quadriplegia | IV; II; II; I; II | ► Medical disorders: Scoliosis, eating difficulties<br>► Neurological disorders: Epilepsy<br>► Mental/behavioural disorders: Mild intellectual disability, autism<br>► Other interventions/medical events during study: Nil | Breaststroke (no kick) | S3/SB2/SM3 |
| 2 | 15 years, 7 months | 19 years, 5 months | Female | Mixed spastic/ dystonic Quadriplegia | IV; II; III; I; I | ► Medical disorders: Gastro-oesophageal reflux disease, profound hearing impairment, shoulder impingement syndrome.<br>► Neurological disorders: Epilepsy<br>► Mental/behavioural disorders: Mild intellectual disability<br>► Other interventions/medical events during study: Baclofen pump malfunction (x2), acute mental health episode (10 weeks). | Backstroke (no kick) | S2/SB2/SM2 |
| 3 | 15 years, 7 months | 19 years, 5 months | Male | Spastic Quadriplegia | IV; II; I; I; II | ► Medical disorders: eating difficulties.<br>► Neurological disorders: Nil<br>► Mental/behavioural disorders: Anxiety<br>► Other interventions/medical events during study: Surgical hallux valgus correction | Backstroke (no kick) | S2/SB2/SM2 |

*Gross Motor Function Classification System.
†Manual Ability Classification System.
‡Communication Function Classification System.
§Visual Function Classification System.
¶Eating and Drinking Classification System.
**Categorised using descriptions from Hollung.[13]

(16-month MB-SCED and 30-month follow-up period) provided a basis for comparing participant motor function with predicted motor decline.[2] During the 30-month follow-up period, the training phases were extended from 16 weeks to longer training blocks aligning with the competitive swimming season, and the withdrawal phases were incorporated between seasons to facilitate recovery. Gross motor function, swimming performance and training load were longitudinally monitored.

## Intervention

The intervention comprised performance-focused swimming training over the course of four consecutive competitive swimming seasons (one Paralympic cycle). The term 'performance-focused' refers to the fact that the sole aim of all strategies employed was to improve competitive swimming performance over 50 m. A comprehensive description of the training programme is available in the published protocol.[10] Training aimed to achieve three main goals:

1. Improve water safety skills.
2. Minimise hydrodynamic drag forces.
3. Maximise propulsive forces.

Training was delivered by a multiprofessional team comprising qualified physiotherapists, exercise physiologists and swim coaches, supported by a multiprofessional medical team. Training session frequency increased from once per week to five times per week as the training phases progressed. Training session intensity and duration varied but aimed to gradually increase over time. The participants were paired with a typically developing volunteer training buddy who provided training assistance.

## Outcomes

In the MB-SCED, repeated measures of swimming performance and gross motor function were conducted throughout five phases: A1 (Baseline)-B1-A2-B2-A3 with staggered exposure/withdrawal sequences.[10] In accordance with SCED guidelines,[19] a minimum of five data points occurred for each participant in each phase.[10] During the baseline phase, participant 1 completed 5 data points, participant 2 completed 8 data points and participant 3 completed 11 data points. All participants then completed: phase B1 (8 data points), phase A2 (5 data points), phase B2 (8 data points) and phase A3 (5 data points).[10]

### Swimming velocity

A full description of the test protocol, including rationale, is reported in the published prototol.[10] To summarise, each participant completed a maximum-effort swimming trial. The duration of each participant's test was based on the 2017 World Para Swimming Championships 50 m freestyle qualifying time for the participant's class, and they swam their preferred stroke as fast and as far as possible in this allotted time. The distance covered was recorded and average swimming velocity was calculated.

### Gross motor function

The Gross Motor Function Measure-66-Item Set (GMFM-66-IS) has excellent levels of overall agreement with the full version of the GMFM-66 when measuring change over time (Intraclass Correlation Coefficient or ICC≥0.9).[20] Given the time-intensive nature of the full test, the short item set was appropriate for use in this study as a repeated measure of gross motor function. Scores for the tasks within the item set were entered into the Gross Motor Ability Estimator programme to obtain the final GMFM-66-item score.

### Training load

Training load comprised the frequency (training sessions per week), duration (minutes spent training) and intensity, which in this study was quantified using the session-RPE (rating of perceived exertion) method.[21] Each participant rated each training session intensity on the OMNI RPE scale[22] which ranges from 0 (extremely easy) to 10 (extremely hard), and this rating was multiplied by the session duration to produce a given number of session RPE minutes. Weekly totals for RPE minutes were calculated.

## Randomisation/blinding

Assessments were conducted by a physiotherapist with expertise in the assessment of gross motor function in people with CP. The assessor was blinded to the intervention and whether each participant was in a period of training or withdrawal at the time of assessment. Participants were randomised to either a 10-week (5 data point), 16-week (8 data point) or 22-week (11 data point) baseline period.

## Statistical methods

Interrupted time-series simulation (ITSSIM) analysis[23] was used to calculate a standardised mean difference effect size, d, and an unstandardised mean difference, D, for each participant's transition from A-B or B-A in the outcomes of swimming performance and GMFM-66. Standardised effect sizes were interpreted as follows: small, 0.20–0.49; moderate, 0.50–0.80 and large, greater than 0.80.[24] The 5-phase design comprised a total of 12 transitions—4 transitions (from an A-B or B-A phase) for each of three participants. The criterion for inferring causality was statistically significant effects for at least three transitions[25]).

The longitudinal non-linear fluctuations in GMFM, as a function of participant age, were evaluated using a generalised additive model with a penalised cubic regression spline basis function and visualised using the 'ggplot' function from the 'ggplot2' package (R Studio V.1.3.1056, PBC, Boston, Massachusetts, USA). A Gaussian distribution with an identity link function was used to produce the general additive model. Five knots were included in the model, positioned at quartiles of the observed data points. The fitted smoothed coefficients resulting from the analysis were plotted along with the 95% CIs. The GMFM fluctuations that could be expected to occur in people who are of the same age as those in the current study were plotted according to the original models developed by Hanna *et al*.[2]

## Equity, diversity and inclusion and patient involvement statement

Equity and patient voice were fundamental to our justification for this study and at the forefront of the discussion of results, implications for future research and clinical practice. This work is driven by the voices of people with disabilities who have high support needs and who acted as consumer advisors for the Para-START programme of research. Our research team comprises both males and females from three countries and includes senior, mid-career and early-career academics.

# RESULTS

## MB-SCED to evaluate the effects of a performance-focused swimming programme

Figure 1 presents training load, GMFM-66 and swimming performance data for each participant over the five phases of the 16-month SCED study. Training load is presented graphically in the three panels on the left side of figure 1. It shows that, in accordance with a multiple baseline design, the baseline (phase A1) is 11, 17 and 23 weeks for participants 1, 2 and 3, respectively. Training load during baseline and the two withdrawal periods (A2 and A3) was zero. Table 2 presents an overview of the training load completed in each of the two training phases phase B1 and B2. Total RPE minutes accrued during B1 were 10888, 7572 and 11 028 for participants 1, 2 and 3, respectively. Total RPE minutes accrued during B2 were 14673, 10 008 and 11 164, respectively.

The three middle panels of figure 1 present swimming velocity, with each participant achieving greater swimming velocity in each training phase. Table 3 presents the results of the ITSSIM analysis for swimming velocity. Data are presented for each participant and each phase transition. All participants achieved

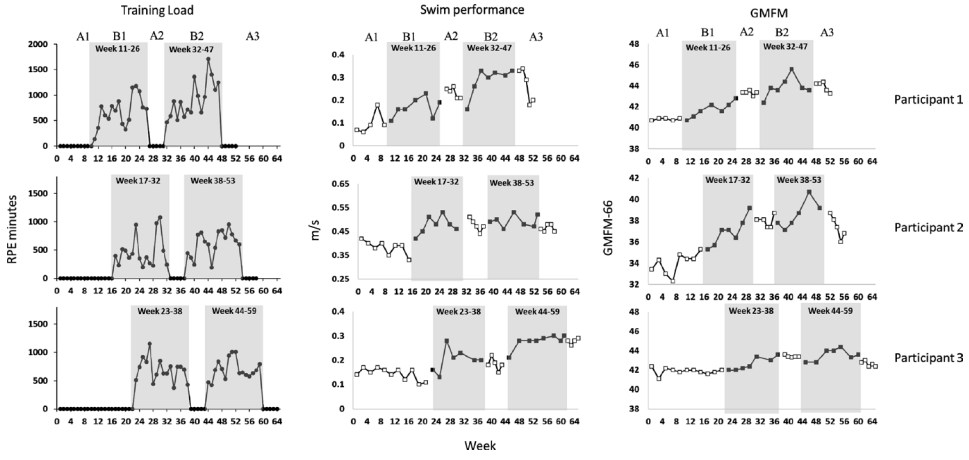


**Figure 1** Training load, swimming performance and GMFM-66 data for each participant throughout the five-phase A1-B1-A2-B2-A3 SCED study. GMFM-66, Gross Motor Function Measure-66-Item Set; SCED, Single-Case Experimental Design.

Table 2 Training load data for each participant presented by training phase

| | Participant 1 | | Participant 2 | | Participant 3 | |
|---|---|---|---|---|---|---|
| Training load measure | Phase B1 | Phase B2 | Phase B1 | Phase B2 | Phase B1 | Phase B2 |
| Total RPE minute | 10 888 | 14 673 | 7572 | 10 008 | 11 028 | 11 164 |
| Mean RPE min/week (SD)* | 681 (298) | 917 (363) | 473 (282) | 625 (222) | 689 (202) | 698 (178) |
| Total distance in metres | 6285 | 12 915 | 3590 | 5570 | 5200 | 6702 |
| Mean distance/week in metres (SD) | 403 (95) | 808 (160) | 224 (71) | 348 (82) | 325 (89) | 414 (68) |
| Total number of training sessions | 33 | 52 | 27 | 36 | 32 | 38 |
| Median number of training session/week (IQR) | 2 (1) | 3 (2) | 1 (1) | 2 (1) | 2 (0) | 2 (1) |

*750 RPE min/week is a volume equivalent to the WHO physical activity recommendation of 150 min of moderate intensity physical activity per week.[26]
RPE, rating of perceived exertion.

increases in swimming velocity each time the intervention was introduced (transitions A1-B1 and A2-B2), and effect sizes were moderate-large (0.61–3.75). In one instance—participant 3, transition A2-B2—the increase in swimming velocity was not statistically significant (p=0.11). Responses to withdrawal of the intervention (transitions B1-A2 and B2-A3) were more variable. Swimming performance in participants 2 and 3 decreased in the B1-A2 transition and effect sizes were moderate-large (−0.69 to −1.82); swimming velocity increased in participant 1 in this transition, though the effect size was small (0.39). Swimming velocity in participants 1 and 3 decreased in the B2-A3 transition and effect sizes were large (−1.17 to −1.64); swimming velocity increased in participant 2 in this transition, though the effect size was small (0.29).

GMFM-66 scores are presented in the three right panels of figure 1 and the results of the ITSSIM analysis are presented in table 3. All participants achieved increases in GMFM-66 score each time the intervention was introduced (transitions A1-B1 and A2-B2). Effect sizes were large in transition A1-B1 (1.15–2.26), but small-moderate in transition A2-B2 (0.11–0.74). GMFM-66 score decreased in all participants each time the intervention was withdrawn (transitions B1-A2 and B2-A3). Effect sizes were moderate-large in transition B1-A2 (−0.50 to −2.01) and small-large in transition B2-A3 (−0.47 to −2.28).

### Comparison of measured and predicted motor function over 46 months

The raw weekly training load and modelled GMFM-66 data for each participant over 46 months are presented in figure 2. Training load remains relatively consistent over the entire period, although participant 1 has some large peaks in the third training period (aged 17 years). The red line indicates 750 RPE min/week, the volume of activity recommended for people with disabilities by the WHO.[26]

The three right-hand panels of figure 2 present modelled GMFM-66 data for each participant. Scores increase in the first year of training (age 15–16 years), and then plateau in the subsequent 3 years into late adolescence. The red line in each GMFM panel is the predicted trajectory for GMFM-66 scores.[2] For each participant, the red line originates from the median GMFM-66 score at baseline for each participant. The upward trend of modelled GMFM-66 measures for each participant contrasts with the predicted downward trend in GMFM-66 indicated by the red line.

## DISCUSSION

There were two main findings from this study. First, a performance-focused swimming training programme comprising training volumes commensurate with WHO physical activity recommendations and delivered by a skilled multiprofessional team conferred improved motor function in previously untrained, physically inactive people with CPHSN. The five-phase SCED demonstrated that motor function improved following training phases and declined following withdrawal phases in all participants, thereby indicating the relationship was causal—performance-focused swimming caused gross motor function to improve.

The second main finding was that, over a 46-month period, participant gross motor function initially improved and then plateaued around the new, improved level. These improvements occurred during a life stage when population-based modelling[2] indicates that motor function typically declines. Specifically, the participants were aged 15/16 years at baseline and their GMFM-66 scores improved by between 2 and 7 points from their median baseline score and then plateaued until age 19/20 years. During the same life stage, population-based modelling predicts mean GMFM-66 scores typically fall by 4.2 points for people with GMFCS level IV CP.[2] Thus, the difference between predicted and measured motor function for participants in this project was between 6.2 and 11.2 points on the GMFM-66 scale, a clinically meaningful difference. The plateau in motor function indicated a ceiling effect—participants may have, at least to some extent, maximised their gross motor capacity as measured using the GMFM-66.

Together, this study's two main findings indicate that people with CP at GMFCS level IV who achieve physical activity guidelines during adolescence may not only prevent motor decline but improve it. The obverse of this finding is that the high prevalence of physical inactivity in this group during adolescence may account for declines which are currently accepted as clinically inevitable. This may have implications for clinical practice—and highlights the importance of including physical activity interventions as part of routine care of adolescents with CP.

Improvement in swimming velocity for all three participants validated our characterisation of the training programme as 'performance focused'. Results support the veracity of previously reported athlete testimonies which claim that meaningful improvements in physical function are conferred by performance-focused sports training.[9]

We suggest three key features of the programme contributed to the results observed:

1. The competitive sport context: For young people with CPHSN, competitive sport has a number of advantages, and the views of ParaSTART participants have been published.[27] In addition, competitive sport is age appropriate and culturally significant for many young people with CP; is routinely supported by multiprofessional teams; focuses on achievement of excellence, rather than identifying and remediating

**Table 3** Swimming performance and GMFM-66-IS ITSSIM results

| Outcome measure | Participant | A1-B1 | | | | B1-A2 | | | A2-B2 | | | | B2-A3 | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | Null effect mean (SE) | Exp effect mean (SE) | D | D (SE) | P value | Null effect mean (SE) | Exp effect mean (SE) | D | D (SE) | P value | Null effect mean (SE) | Exp effect mean (SE) | D | D (SE) | P value | Null effect mean (SE) | Exp effect mean (SE) | D | D (SE) | P value |
| Swimming velocity | 1 | 0.15 (0.05) | 0.18 (0.05) | 0.03 | 0.61 (0.62) | <0.01 | 0.21 (0.10) | 0.24 (0.02) | 0.03 | 0.39 (0.61) | 0.06 | 0.18 (0.05) | 0.31 (0.04) | 0.13 | 2.78 (0.82) | 0.001 | 0.43 (0.11) | 0.29 (0.04) | −0.14 | −1.64 (0.67) | <0.001 |
| | 2 | 0.34 (0.05) | 0.48 (0.03) | 0.14 | 3.75 (0.83) | <0.001 | 0.57 (0.07) | 0.47 (0.02) | −0.10 | −1.82 (0.67) | <0.001 | 0.36 (0.05) | 0.49 (0.03) | 0.13 | 2.92 (0.87) | <0.001 | 0.46 (0.08) | 0.48 (0.03) | 0.02 | 0.29 (0.61) | <0.001 |
| | 3 | 0.12 (0.03) | 0.20 (0.04) | 0.08 | 2.35 (0.62) | <0.001 | 0.24 (0.11) | 0.18 (0.03) | −0.06 | −0.69 (0.6) | <0.001 | 0.14 (0.09) | 0.28 (0.03) | 0.14 | 2.33 (0.78) | 0.11 | 0.34 (0.06) | 0.28 (0.02) | −0.06 | −1.77 (0.66) | <0.001 |
| GMFM-66 | 1 | 40.90 (0.37) | 41.60 (0.31) | 0.70 | 2.11 (0.73) | <0.001 | 43.64 (0.58) | 43.40 (0.28) | −0.24 | −0.5 (0.59) | <0.001 | 43.40 (0.75) | 43.80 (0.92) | 0.40 | 0.47 (0.59) | <0.001 | 45.00 (2.19) | 44.20 (0.39) | −0.8 | −0.47 (0.59) | <0.001 |
| | 2 | 35.72 (1.50) | 37.10 (0.68) | 1.38 | 1.15 (0.56) | <0.001 | 40.25 (1.25) | 38.10 (0.72) | −2.15 | −2.01 (0.72) | <0.001 | 38.10 (1.73) | 38.26 (1.06) | 0.16 | 0.11 (0.61) | <0.001 | 41.13 (2.12) | 37.40 (0.67) | −3.73 | −2.28 (0.78) | <0.001 |
| | 3 | 41.64 (0.39) | 42.40 (0.24) | 0.76 | 2.26 (0.61) | <0.001 | 44.00 (0.51) | 43.40 (0.13) | −0.60 | −1.49 (0.66) | <0.001 | 43.25 (0.38) | 43.60 (0.53) | 0.35 | 0.74 (0.61) | <0.001 | 44.40 (1.19) | 42.60 (0.25) | −1.8 | −1.93 (0.71) | <0.001 |

GMFM-66, Gross Motor Function Measure-66-Item Set; ITSSIM, interrupted time-series simulation.

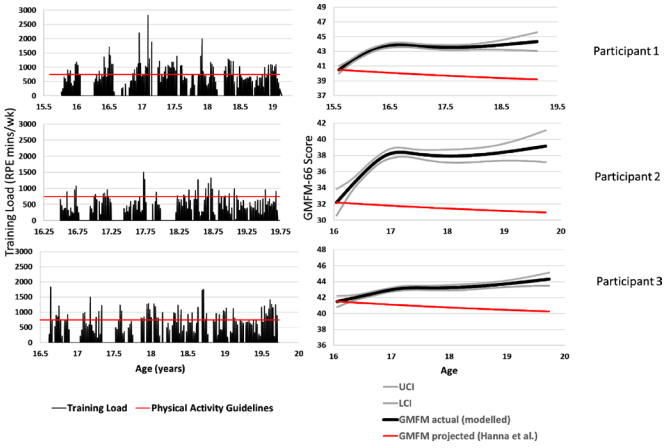


**Figure 2** Longitudinal training load and GMFM-66 data. The left panel displays training load data for each participant between the age of 15/16 years and 19/20 years (displayed x-axis; note that the baseline period is not temporally represented). The red horizontal line denotes the RPE-minute value commensurate with national physical activity guidelines (750 RPE min/week). The right panel displays modelled GMFM data for each participant, with 95% CIs, and the red line denotes the projected trajectory of motor decline,[2] from the median of baseline GMFM-66 scores. GMFM-66, Gross Motor Function Measure-66-Item Set; LCI, lower confidence interval; RPE, rating of perceived exertion; UCI, upper confidence interval.

motor-sensory impairments and fosters personal interaction and teamwork[28]—critical features for youth who often experience social isolation and challenging periods of transition between adolescence and adulthood.[29]

2. Qualified multiprofessional staff: The delivery team included physiotherapists, exercise physiologists and coaches. They were supported by a medical doctor, dietician, occupational therapist, speech pathologist and sport psychologist. Heterogeneous, complex comorbdities and medical events/issues were managed during the programme: the number of comorbidities/medical issues for participants 1, 2 and 3 were N=5; N=8 and N=3, respectively. Table 1 lists the comorbidities and medical issues of each participant. Participants in this study could not be safely, effectively accommodated in a non-specialist, community-based swimming club.
3. Transport costs supported: Participants were not independent on public transport and required either a taxi or family member to drive them. Associated expenses were met by research funding and community donations.

The importance of this study is amplified because little is known about exercise training responses in people with CP, GMFCS level IV.[7] In the absence of research evidence, some clinicians and researchers have vastly underestimated the physical capabilities of this group. One recent review stated that people at GMFCS IV and V '…will struggle performing structured exercise programmes' and 'are unable to perform activities greater than 1.0 MET'.[30] Note that 1.0 METs is the energy expended during quiet sitting.[31] Low rates of physical activity participation and gross under-representation in exercise training trials[7] may result from such assertions and are refuted by results from this study. Future studies should include those with CPHSN.

Methodologically, the SCED used was ideally suited to the study aims. The design generated high-level evidence[14] and conferred a range of advantages[10] including permitting the allocation of time and expertise required to safely supervise participants with severe primary impairments and multiple

comorbidities who were at increased risk of serious adverse events (see table 1); providing personalised assistance to alleviate the increased time cost associated with training[12] and providing the methodological freedom to individualise training type, duration and intensity without compromising experimental control.

Importantly, the SCED overcame the arguably impossible task of achieving both adequate sample size and satisfactory participant homogeneity in relation to key prognostic variables for a group-level study design. Specifically, we posit that the absence of RCTs investigating responses to sport and exercise training interventions in people with CPHSN may be due, at least in part, to the infeasibility of recruiting a sample that is both large enough to adequately power the trial and also sufficiently homogenous with respect to key prognostic variables (age, sex, neurological subtype, functional effects and comorbidities). Wider use of the SCED may facilitate generation of high-quality Para sport and exercise training evidence in other heterogeneous populations, including people with acquired brain injuries and spinal cord injuries.

## Limitations

This study has several limitations. First, the small number of participants and use of the SCED enhanced internal validity in this study but limited external validity. This necessitates cautious interpretation of the generalisability of the results. Second, the age range within the sample was narrow (all aged 15 years on enrolment), and it is possible that children of different ages may respond differently. Further longitudinal studies throughout the known period of decline (from age 7 years to 21 years) are required. Finally, free-living physical activity was not measured during the baseline or withdrawal periods. Although people with CPHSN typically accumulate low volumes of daily activity[6] and no training was conducted during these periods, we did not control for this effect.

## CONCLUSION

This study demonstrated that performance-focused swimming training provided a context for adolescents with CPHSN to accumulate health-enhancing volumes of physical activity, improve their swimming performance and their gross motor function during a life stage when population-based modelling predicts gross motor decline. However, this is a clinically complex population. In order to permit their effective participation in sports, priority should be placed on the development of procedures and programmes that can be delivered by a multiprofessional team. Further research employing SCED methodology is required, with emphasis on replication in this population and in other Para sports.

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**Competing interests** None declared.

**Patient and public involvement** Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

**Patient consent for publication** Consent obtained directly from patient(s).

**Ethics approval** This study involved human participants and two ethical approvals were provided by the University of Queensland Ethics Committee: UQ approval #2015000831, which applied to the initial 16-month MB-SCED; and UQ approval #2018001472 which applied to the 30-month follow-up study. Participants gave informed consent to participate in the study before taking part.

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## REFERENCES

1 Bax M, Goldstein M, Rosenbaum P, *et al*. Proposed definition and classification of cerebral palsy, April 2005. *Dev Med Child Neurol* 2005;47:571–6.
2 Hanna SE, Rosenbaum PL, Bartlett DJ, *et al*. Stability and decline in gross motor function among children and youth with cerebral palsy aged 2 to 21 years. *Develop Med Child Neuro* 2009;51:295–302.
3 Smith SE, Gannotti M, Hurvitz EA, *et al*. Adults with cerebral palsy require ongoing neurologic care: a systematic review. *Ann Neurol* 2021;89:860–71.
4 Gormley ME, Deshpande S. Hypertonia. In: Murphy KP, Houtrow AJ, eds. *Pediatric rehabilitation: principles and practice*. New York: Springer Publishing, 2020.
5 Ryan JM, Forde C, Hussey JM, *et al*. Comparison of patterns of physical activity and sedentary behavior between children with cerebral palsy and children with typical development. *Phys Ther* 2015;95:1609–16.
6 Verschuren O, Peterson MD, Balemans ACJ, *et al*. Exercise and physical activity recommendations for people with cerebral palsy. *Dev Med Child Neurol* 2016;58:798–808.
7 Lai B, Lee E, Kim Y, *et al*. Leisure-time physical activity interventions for children and adults with cerebral palsy: a scoping review. *Develop Med Child Neuro* 2021;63:162–71.
8 Clutterbuck G, Auld M, Johnston L. Active exercise interventions improve gross motor function of ambulant/semi-ambulant children with cerebral palsy: a systematic review. *Disabil Rehabil* 2019;41:1131–51.
9 Tweedy SM, Beckman EM, Johnston LM, *et al*. Performance-focussed sport – an avenue to gold-medal clinical outcomes for people with neurological impairments? *Brain Impairment* 2016;17:99–110.
10 Dutia IM, Connick MJ, Beckman EM, *et al*. Evaluating the effects of performance-focused swimming training on people with cerebral palsy who have high support needs - a study protocol using single-case experimental design. *Brain Impairment* 2020;21:217–34.
11 Dimitrijević L, Aleksandrović M, Madić D, *et al*. The effect of aquatic intervention on the gross motor function and aquatic skills in children with cerebral palsy. *J Hum Kinet* 2012;32:167–74.
12 Dutia I, Curran D, Donohoe A, *et al*. Time cost associated with sports participation for athletes with high support needs: a time-motion analysis of tasks required for para swimming. *BMJ Open Sport Exerc Med* 2022;8:e001418.
13 Hollung SJ, Bakken IJ, Vik T, *et al*. Comorbidities in cerebral palsy: a patient Registry study. *Develop Med Child Neuro* 2020;62:97–103.
14 Nikles J, Mitchell G. The essential guide to N-Of-1 trials in health. In: *SpringerLink content*. 1st edn. Dordrecht: Springer, 2015.
15 OCEBM Levels of Evidence Working Group. *The Oxford 2011 levels of evidence*. 2011.

16 McDonald S, Quinn F, Vieira R, *et al*. The state of the art and future opportunities for using longitudinal N-Of-1 methods in health behaviour research: a systematic literature overview. *Health Psychol Rev* 2017;11:307–23.
17 Tweedy SM, Connick MJ, Beckman EM. Applying scientific principles to enhance paralympic classification now and in the future: a research primer for rehabilitation specialists. *Phys Med Rehabil Clin N Am* 2018;29:313–32.
18 Zacca R, Azevedo R, Chainok P, *et al*. Monitoring age-group swimmers over a training macrocycle: energetics, technique, and anthropometrics. *J Strength Cond Res* 2020;34:818–27.
19 Logan LR, Hickman RR, Harris SR, *et al*. Single-subject research design: recommendations for levels of evidence and quality rating. *Develop Med Child Neuro* 2008;50:99–103.
20 Avery LM, Russell DJ, Rosenbaum PL. Criterion validity of the GMFM-66 item set and the GMFM-66 basal and ceiling approaches for estimating GMFM-66 scores. *Dev Med Child Neurol* 2013;55:534–8.
21 Wallace LK, Slattery KM, Coutts AJ. The ecological validity and application of the session-RPE method for quantifying training loads in swimming. *J Strength Cond Res* 2009;23:33–8.
22 Fragala-Pinkham M, O'Neil ME, Lennon N, *et al*. Validity of the OMNI rating of perceived exertion scale for children and adolescents with cerebral palsy. *Develop Med Child Neuro* 2015;57:748–53.
23 Tarlow KR, Brossart DF. A comprehensive method of single-case data analysis: interrupted time-series simulation (ITSSIM). *School Psychology Quarterly* 2018;33:590–603.
24 Cohen J. *Statistical power analysis for the behavioural sciences*. 2nd edn. Hillsdale, NJ: Lawrence Erlbaum Associates Inc, 1988.
25 Shadish WR, Sullivan KJ. Characteristics of single-case designs used to assess intervention effects in 2008. *Behav Res* 2011;43:971–80.
26 Carty C, van der Ploeg HP, Biddle SJH, *et al*. The first global physical activity and sedentary behavior guidelines for people living with disability. *J Phys Act Health* 2021;18:86–93.
27 Enright E, Beckman EM, Connick MJ, *et al*. Competitive sport, therapy, and physical education: voices of young people with cerebral palsy who have high support needs. *Br J Sports Med* 2021;55:524–5.
28 Aitchison B, Rushton AB, Martin P, *et al*. The experiences and perceived health benefits of individuals with a disability participating in sport: a systematic review and narrative synthesis. *Disability and Health Journal* 2022;15:101164.
29 Bagatell N, Chan D, Rauch KK, *et al*. "Thrust into adulthood": transition experiences of young adults with cerebral palsy. *Disabil Health J* 2017;10:80–6.
30 Toldi J, Escobar J, Brown A. Cerebral palsy: sport and exercise considerations. *Curr Sports Med Rep* 2021;20:19–25.
31 Kenney WL, Wilmore JH, Costill DL. *Physiology of sport and exercise*. 8th edn. Champaign, IL: Human Kinetic, 2021.