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The influence of lower limb impairments on RaceRunning performance in athletes with hypertonia, ataxia or athetosis.

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Conflict of interest

Cerebral Palsy International Sport and Recreation Association (CPISRA) provided funding for Travel and accommodation costs associated with the data collection. None of the authors declare any other conflict of interest.

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Abstract

Objectives RaceRunning enables athletes with limited or no walking ability to propel themselves independently using a three-wheeled running bike that has a saddle and a chest plate for support but no pedals. For RaceRunning to be included as a para-athletics event, an evidence-based classification system is required. Therefore, the aim of this study was to assess the association between a range of impairment measures and RaceRunning performance.

Methods The following impairment measures were recorded: lower limb muscle strength assessed using Manual Muscle Testing (MMT), selective voluntary motor control assessed using the Selective Control Assessment of the Lower Extremity (SCALE), spasticity recorded using both the Australian Spasticity Assessment Score (ASAS) and Modified Ashworth Scale (MAS), passive range of motion (ROM) of the lower extremities and the maximum static step length achieved on a stationary bike (MSSL). Associations between impairment measures and 100-meter race speed were assessed using Spearman's correlation coefficients.

Results Sixteen male and fifteen female athletes (27 with cerebral palsy), aged 23 (SD=7) years, Gross Motor Function Classification System ranging from II to V, participated. The MSSL averaged over both legs and the ASAS, MAS, SCALE, and MMT summed over all joints and both legs, significantly correlated with 100 m race performance (rho: 0.40-0.54). Passive knee extension was the only ROM measure that was significantly associated with race speed (rho=0.48).

Conclusion

These results suggest that lower limb spasticity, isometric leg strength, selective voluntary motor control and passive knee extension impact performance in RaceRunning athletes. This supports the potential use of these measures in a future evidence-based classification system.

Keywords: RaceRunning, running, Cerebral Palsy, para-athletics, spasticity, selective motor control.

INTRODUCTION

In the Paralympics, people with neurological conditions leading to hypertonia, ataxia and athetosis such as cerebral palsy (CP) are eligible to compete in a number of sports. However, those with total body involvement are often limited to sports with relatively low levels of aerobic and weight bearing demands such as boccia. Low levels of aerobic fitness impose future health risks such as cardiovascular and metabolic diseases [1]. Ryan and colleagues [2] reported that young adults with CP participate less in moderate to vigorous physical activity compared to typically developed peers, increasing their risk of developing cardio-metabolic disease. Further, there is evidence that those with CP who are non-ambulant are more prone to complications from not weight bearing such as lower limb muscle weakness [3] and reduced bone density [4].

RaceRunning allows people who are not able to functionally run and may have limited or no ability to walk independently, to engage in a sport where they can propel themselves using a running bike or 'RaceRunner'. The running bike has a three-wheeled frame that supports the athlete by way of a saddle and anterior trunk support plate (Fig 1). The athletes propel themselves using their legs and steer using handlebars. As such, RaceRunning allows people whose walking ability is moderately to severely impaired, to take part in an aerobic activity. Bolster et al [5] reported maximum heart rates ranging from 168 to 174 beats/min in a study assessing the reliability of the 6 minute Racerunner test. Assuming a maximum heart rate of around 194 bpm in young people with CP [6], these heart rates indicate that the participants (GMFCS levels III and IV) were exercising at heart rates well above 55% of their maximum heart rate recommended by the American College of Sports Medicine for moderate to vigorous physical activity.

Currently, RaceRunning is governed by the Cerebral Palsy International Sport and Recreation Association (CPISRA). For athletes with a neurological condition resulting in hypertonia, ataxia and/or athetosis there are currently three main classes; RR1, RR2 and RR3. Athletes in RR1 are those with the highest activity limitation. CPISRA is currently developing RaceRunning for inclusion in future Paralympic Games as a para-athletics event. In para-athletics, people with hypertonia, ataxia and/or athetosis compete in one of eight classes; four for ambulant athletes (T35-T38) and four for wheelchair athletes (T31-T34). However, at the 2016 Paralympic games there were no track events for those with total body involvement, i.e. classes T31 and T32.

All Paralympic events are required to have an evidence-based classification system that promotes participation by minimising the impact the athlete's impairment on the outcome of the competition [7]. Key components in classification research are the selection of impairment measures and the determination of the strength of association of these measures with performance or performance indicators. Relatively few studies assessed the association of impairment measures with sports performance in athletes with hypertonia, ataxia and/or athetosis [8.9] and no studies have yet been published on the relationship between impairment measures and performance in RaceRunning. However, the impact of lower limb impairments, such as muscle strength, spasticity, range of motion and selective motor control on walking performance in young people with CP has received much attention in the rehabilitation literature (e.g. [10-14]). The evidence presented in these studies, combined with the current CPISRA classification of RaceRunning resulted in the selection of several measures of lower limb impairments; spasticity, passive range of motion, manual muscle strength and selective voluntary motor control (SVMC). These measures were hypothesised to be associated with RaceRunning performance and are thought to be relatively resistant to the effect of RaceRunning training. We also included a test that assessed sport-specific impairment: the maximum step length that could be achieved while seated on a stationary running bike.

The aim of this study was to assess the association of a range of measures with RaceRunning performance during competition. This study focused on the 100m sprint performance, as this is the most popular event amongst RaceRunning athletes at the annual International RaceRunning Cup. We hypothesised that the selected impairment measures would show moderate (Spearman's rho 0.3-0.5) correlations with RaceRunning speed over 100m.

METHODS

Participants and Recruitment

Female and male athletes with hypertonia, ataxia, athetosis or a combination of these, who are in RaceRunning classes RR1/ RR1 assist, RR2 or RR3, aged between 14 and 45 and with at least one year experience of RaceRunning were eligible for inclusion. Potential participants were recruited from RaceRunning clubs in Scotland and from athletes taking part in the international RaceRunning Camp & Cup, which included the CPISRA European Championships in 2016. The study was approved by the Queen Margaret University Research Ethics Committee. All participants gave informed consent or where appropriate, assent prior to taking part. For those under 16, parental consent was obtained.

Descriptive measures

The following data was also collected: Gender, age, Gross Motor Function Classification System (GMFCS) level [15], Functional Mobility Scale [16], years of RaceRunning experience, leg length and CPISRA RaceRunning classification. Standing and walking ability was assessed by two Gross Motor Function Measure (GMFM-66) items; free standing for 20 seconds (item 56), and walking forward 10 steps unsupported (item 69). The scoring for these items ranges from 0 to 3, where 0 indicates the movement has not been initiated and 3 indicates the task was completed [17].

Potential classification measures

Spasticity, isometric muscle strength, SVMC and passive range motion were assessed by an experienced specialist neuro-paediatric physiotherapist who is also a para-athletics classifier (NT) with the assistance of a physiotherapy student (SJ).

Spasticity

Spasticity of the hip adductors, hamstrings, quadriceps and the gastrocnemius were measured in both legs using two different scales; the Modified Ashworth Scale (MAS) [18] and the more recently developed Australian Spasticity Assessment Scale (ASAS). The ASAS [19] is the scale used in the current CPISRA RaceRunning classification system while the IPC classification systems use the MAS. The inter-rater reliability of the ASAS in children with CP has been reported as good (kappa: 0.87) (25). Both measures were included to assess which one would result in the strongest association with running speed.

Isometric muscle strength

Lack of muscle strength of the hip abductors, hip and knee extensors and hip flexors was evaluated manually. The method proposed by Daniels and Worthington [20] was adapted by taking into account the passive range of motion of the muscle group. People with CP are often unable to achieve normal range of motion due to increased tone or contractures but have normal or near normal strength in mid or outer range. Therefore, first the passive range of motion of the muscle group was assessed and ranked as either outer, mid or inner range. This ranking was achieved by dividing the full physiological range of motion in three equal ranges, with outer range defined as the most severely restricted range of motion, inner range as the least restricted (near) normal range and the mid range in between the outer and inner range.

Following the estimation of the passive range of motion, the level of muscle weakness using the 0-5 scoring was rated in that particular range. A score of Inner 5 indicates normal strength in inner (normal) range, while a score of Mid 0 means that the limb moves passively to mid range, but there is no muscle activity in this range. This scoring system allows 18 different scores, ranging from Outer 0 to Inner 5. For the purpose of the analysis, these scores were transformed to an ordinal scale from 0 (Inner 5, no strength impairment) to 17 (Outer 0, no muscle activity in outer range, maximum strength impairment). All tests were performed with the participant either sitting or lying. The inter-rater reliability for the original 0-5 scoring was reported to be moderate to good for children with muscular dystrophy [21].

Selective voluntary motor control

The Selective Control Assessment of the Lower Extremity (SCALE) assesses the ability to perform isolated movements at the hip, knee, ankle, subtalar and toe joints [22]. SVMC was scored as "normal" (2 points) if the athlete could move the tested joint selectively (e.g. without moving other joints), within at least 50% of the possible range of motion and at a physiological cadence cued verbally by the examiner. If any deviation in performance occurred (movement performed slower, below 50% of range of movement, with movement in other joints), selectivity was regarded as "impaired" (1 point). The score "unable" (0 points) was given if no joint movement could be made or only with simultaneously moving other joints. The validity of the SCALE has been established by strong correlations (rho> 0.8) with the Gross Motor Function Classification System [22,23] and the Fugl-Meyer Test (items III-IV) [23] in children with spastic CP. The same studies also demonstrated a high level of interrater-reliability (ICC > 0.8) [22,23] .

Passive range of motion

The following measures of passive range of motion (ROM) were recorded using a clear plastic rigid goniometer to the nearest degree: maximum hip flexion with the knee flexed and with the knee extended, popliteal angle, maximum hip extension, maximum knee extension, maximum knee flexion, maximum dorsiflexion with the knee extended and with the knee flexed and the hindfoot – thigh angle [24]. Kilgour et al [25] reported high Intra-session reliability (ICC > 0.9) for passive range of motion measures in children with CP.

Maximum static step length (MSSL)

In addition to the above impairment measures, a sport specific measure was also included. For this test, athletes took as big a step as possible when seated on their running bike but without rolling forward. Starting position was with the feet parallel to each other vertically under the saddle/hip. The distance between the foremost point of the shoe at the starting position and at the end of the step was measured to the nearest mm for each leg. Intra session reliability assessed for 10 athletes was high (ICCs=0.98 for both the left and right leg).

RaceRunning performance

RaceRunning performance was measured as the fastest 100 meter time achieved during an international RaceRunning event in July 2016 or just before or after. For the correlational analysis, the 100 meter race time was converted into dimensionless running speed to correct for the effect of leg length using the equation proposed by Hof [26]. This correction for leg length removed the confounding effect of leg length when analysing the associations between impairment and running speed. We do not propose that leg length could be included as a classification measure.

Statistical analysis

For the SCALE, ASAS, MAS and Manual Muscle Testing (MMT) measures, the individual joint values were summed for both legs to calculate a total score. The values for each ROM measure and the MSSL were averaged over both legs.

The associations between running speed and the lower limb impairment measures and the MSSL were determined using Spearman rank coefficients. Statistical significance was accepted for $p<0.05$. All statistical analyses were performed using SPSS v19. (IBM statistics, Armonk, USA).

RESULTS

Twenty-nine participants were recruited either in Scotland or during an international RaceRunning event in July 2016. Two further athletes were assessed in February 2017. The majority of the participants had a diagnosis of CP and used a wheelchair or walker to mobilise over a distance of 5 meters (Table 1). Around half of the participants were able to stand unaided for 20 seconds (GMFM item 56) and/or walk 10 steps unaided (GMFM item 67). Average (standard deviation) dimensionless race speeds were 1.35(0.2), 1.49(0.21) and 1.16 (0.22) and 0.94 (0.01) for GMFCS levels II, III, IV and V respectively. Table 2 shows the descriptive statistics of the impairment measures and sport specific measure.

Correlation of impairment measures with running speed

Table 3 shows the associations between running speed and impairment measures of and MSSL. Of the passive ROM measures, only passive hip extension and passive knee extension are included in this table.

None of the other passive ROM measures showed a correlation coefficient higher than 0.3 and none was significantly correlated with running speed.

All impairment measures in table 3, except passive hip extension, were moderately correlated (rho >0.4,) with running speed over 100m. The sport-specific impairment measure, MSSL was also moderately correlated with running speed (rho=0.54).

Figures 2a-2f show the scatter diagrams illustrating the association between measures and RaceRunning speed.

DISCUSSION

This is one of the few studies assessing the extent to which impairments of body function affect performance in para-sport for people with a coordination impairment, and the first study doing this for RaceRunning. The results show that spasticity and lack of selective voluntary motor control, muscle strength and knee range of motion are all associated with RaceRunning speed over 100m (rho > 0.4, p<0.05) with the spasticity measures showing the strongest association.

Interestingly, absolute speed showed slightly higher Spearman coefficients than the dimensionless speed. This could be because either leg length does not greatly influence RaceRunning speed or that some of the participants with data points further away from the regression line (higher speeds than predicted from their impairment levels) were small females, resulting in these 'data points' being even further away from the regression line when corrected for leg length (higher dimensionless speed).

In a sample of 12 male Paralympic (T35, T36, T37, T38) athletes with hypertonia, ataxia and/or athetosis, Connick et al [9] found strong associations between running speed and several functional range of motion measures during the acceleration (0-15m) and maintenance (30-60m) of a 60m maximum sprint

($0.41<r<0.76$). None of their co-ordination measures were associated with sprinting performance ($r \le 0.22$). In the same sample, but investigating the association between isometric strength and sprinting performance measures, Beckman et al [8] reported the highest association for plantar flexor strength ($r=0.33$ and $r=0.43$ for the most and least affected leg respectively), but neither was statistically significant. Neither study assessed spasticity or selective voluntary motor control.

Although there is no such evidence with regard to sports performance of CP athletes, several studies [10-14] have reported an association between impairment measures (e.g. spasticity, muscle strength, passive range of motion, selective voluntary motor control) and gait performance in children with CP. Comparison of the results of these gait studies and our study investigating RaceRunning is however limited. Many of the athletes participating in RaceRunning are not able to walk and have higher GMFCS levels (II-V) than those in the majority of the gait studies, which included mostly participants with GMFCS levels I-III.

None of our range of motion measures, except knee extension, showed a statistically significant association with RaceRunning performance. This may be partly because of the variation in propulsion techniques possible in RaceRunning so that only a severe lack of range of motion is associated with slower RaceRunning speeds. This was illustrated by the scatter diagram of speed vs. knee extension, where only beyond a certain ‘critical’ knee flexion contracture a more pronounced decrease in RaceRunning speed could be observed. It is also possible that measures over more than one joint [9] would result in stronger associations.

GMFCS level may have limited use as a potential classification measure. Although those with GMFCS levels IV and V are clearly slower than the other levels, those with GMFCS levels III were on average faster than those with GMFCS level II.

In addition to the commonly used clinical measures of impaired body function, one sport-specific impairment measure was included; maximum static step length whilst on stationary bike. The results of this measure were promising as its association with dimensionless RaceRunning speed was statistically significant and explained 29% of the variance in running speed. Other important advantages of this measure are that it is ratio-scaled and precise and that it is easy to measure with minimal equipment. However, the resistance to training of MSSL still needs to be established.

Two measures of spasticity were included in this study, but although the ASAS showed consistently stronger associations with running speed, the difference was too small to recommend the use of ASAS instead of the MAS based on these results.

The main limitation of this study is that apart from the ROM measures and MSSL, all measures were ordinal and not ratio-scaled. We also summed the ordinal scores for each muscle group and both legs to calculate total scores. The summation of ordinal sub-scores is not uncommon and used in several validated clinical scales (e.g. [15,27]). Further, we believe that the association between summed clinical scales and performance reported both in our study and others [10,14] supports the use of these summed measures until reliable and clinically valid ratio scales become available. Key to the use of any clinical ordinal scale is a strict standardised protocol such as those of the ASAS [19] and SCALE [19] with proven inter- and intra-reliability and trained, experienced assessors.

Several authors have attempted to quantify spasticity though EMG and torque measures [28,29] or used inertial sensors to more accurately define the point of catch [30]. Unfortunately, these measures are time consuming and require specialist equipment and specialist staff, which makes them largely unsuitable for routine athlete profiling. Futures studies should aim to develop practical, valid, reliable and ratio-scaled measures for this purpose.

The current study did not assess the associations of impairment measures related to trunk control [27] and upper limb function, neither did we measure multi-joint coordination, all of which may be associated with RaceRunning performance. Finally, a relatively low sample size precluded the use of multiple regression analysis.

In conclusion, our study confirmed that spasticity, decreased selective voluntary motor control, impaired muscle strength and the presence of a severe (>20degrees) knee flexion contracture are negatively associated with RaceRunning performance and supports the use of these measures in an evidence based RaceRunning classification system.

Conflict of interest

Cerebral Palsy International Sport and Recreation Association (CPISRA) provided funding for travel and accommodation costs associated with the data collection.

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Cerebral Palsy International Sport and Recreation Association (CPISRA) provided funding for travel and accommodation costs associated with the data collection. CPISRA was not involved in the study design, data collection, analysis, interpretation of data and writing of the manuscript.

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Table 1 Demographics of the participants

	Mean ± SD (range)
Age	23.6 ± 7.01 (14-42)
Gender (male/female)	16/15
100 meter time (s)	28.0 ±6.8 (17.5-46.0)
Years involved in RaceRunning	4.4 (3.0) (1:11)
Diagnosis (CP /TBI/Tumour/NA)	29/1/1
Hypertonia/athetosis/ataxia/mixed/NA	17/1/3/8/2
GMFCS (II/III/IV/V)	8/10/11/2
CPISRA RaceRunning Classification (RR1/RR2/RR3)	11/12/8
FMS 5 meters 1/2/3/4/5/6[¥]	9/9/0/1/12/0
FMS 50 meters 1/2/3/4/5/6[¥]	14/5/0/2/10/0
FMS 500 meters 1/2/3/4/5/6[¥]	18/5/0/1/7/0
GMFM item 56 (ability to stand unaided for 20s): 0/1/2/3	12/2/1/16
GMFM item 69 (ability to walk 10 steps unaided): 0/1/2/3	15/1/0/15

[¥]1: wheelchair, 2: walker or frame 3: crutches, 4: one or two sticks, 5 independent on flat surface, 6: independent on any surface [16]
NA=not available; CP=Cerebral Palsy; TBI= Traumatic Brain Injury; FMS=Functional Mobility Score; GMFCS=Gross Motor Function Classification System, GMFM=Gross Motor Function Measure-66 [17].

Table 2 Impairment and sports specific measures summed (MAS, ASAS, MMT, SCALE) or averaged (knee and hip extension, MSSL) for both left and right leg. For MAS, ASAS and MMT a higher value indicated a higher impairment, for the SCALE and MMSL a higher value indicate a lower impairment.

Measure (range of possible values)	**Mean (SD) [range]**
MAStot (0:32)	13.9(5.8) [0:24]
ASAStot (0:32)	16.8(5.6) [0:24
MMTtot (0:144)	58.3(24.3) [24:108]
SCALEtot (0:20)	12.6(5.7) 4:20
Knee $extension_{av}$ (°)*	-8(14) [-54: 14]
Hip $extension_{av}$(°)*	4(6) [-13 : 19]
$MSSL_{av}$ (cm)	40.4(25.7)[-8:105]

*a negative value indicates flexion contracture

MAS: Modified Ashworth Scale, ASAS: Australian Spasticity Assessment Scale, MMT: Manual Muscle Testing. MSSL: Maximum Static Step Length

Table 3 Spearman rank-correlations between impairment and sports specific measures and dimensionless and absolute running speed over 100m.

	Speed (dimensionless)	**Speed (m/s)**
SCALEtot	.40[*]	.47[¥]
MMTtot	-.47[¥]	-.53[¥]
MAStot	-.48[¥]	-.49[¥]
ASAStot	-.49[¥]	-.51[¥]
Hip Extension	.34	.36
Knee Extension	.48[¥]	.52[¥]
MSSL	54[¥]	.60[¥]

*=p<0.05, ¥=p<0.01

MAS: Modified Ashworth Scale, ASAS: Australian Spasticity Assessment Scale, MMT: Manual Muscle Testing, MSSL: Maximum Static Step Length

Fig 1 RaceRunner (reproduced with permission from Quest88 Ltd)

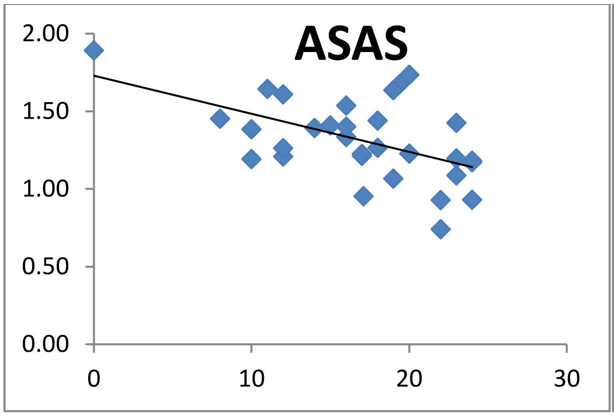

Fig 2a

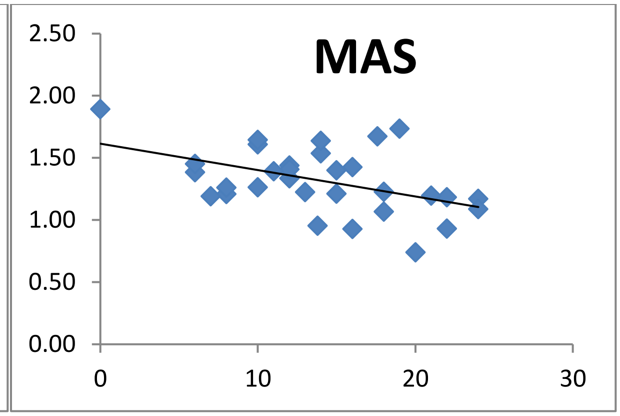

Fig 2b

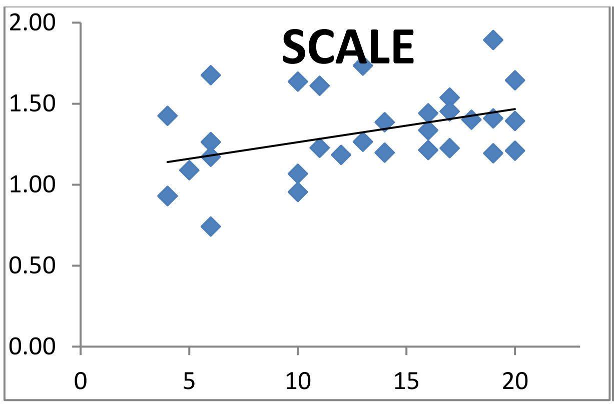

Fig 2c

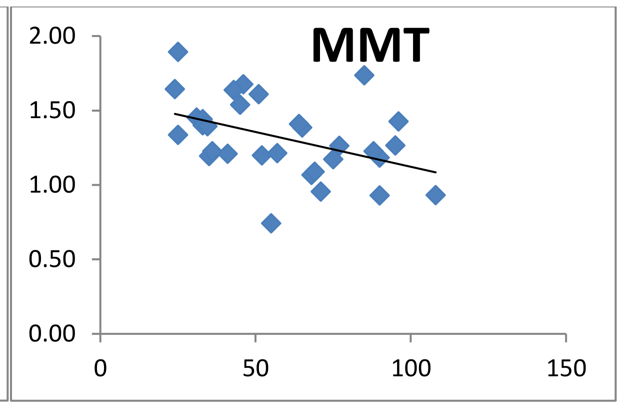

Fig 2d

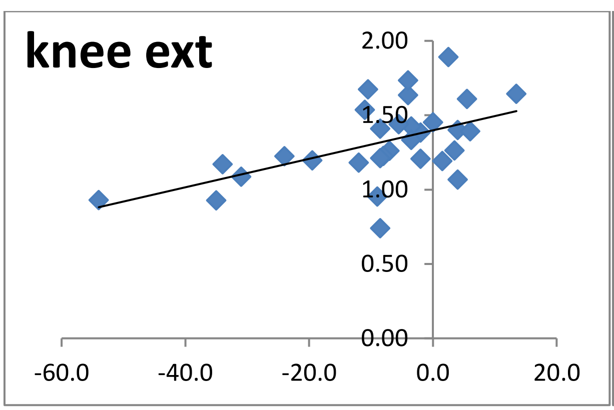

Fig 2e

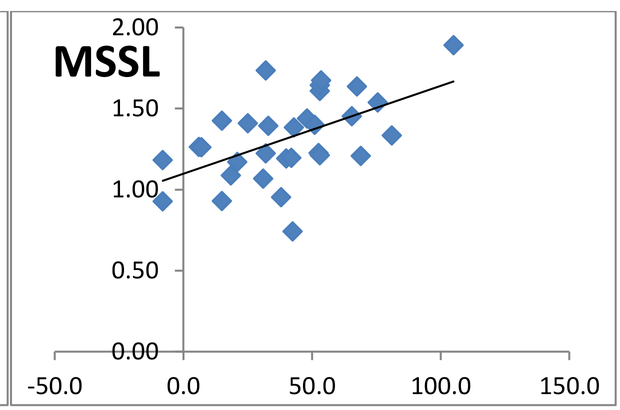

Fig 2f

Fig 2a-2f

Scatter diagrams of the impairments measures versus dimensionless running speed (vertical axis).

For the MAS, ASAS and MMT a higher value indicates a higher impairment, while for SCALE and MSSL a higher value means a lower impairment. Knee flexion contractures are indicated as negative (fig 2e).

Highlights

- RaceRunning allows people with limited walking ability to take part in an aerobic activity.
- Spasticity and muscle weakness are negatively associated with RaceRunning speed.
- Selective voluntary motor control is positively associated with RaceRunning speed.
- A knee flexion contracture is associated with a reduced RaceRunning speed.
- Inclusion of lower limb impairment measures in RaceRunning classification is warranted.