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Clinical use of the modified shuttle test in children with cystic fibrosis: Is one test sufficient?

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Abstract

Objective: To evaluate the reproducibility of the modified shuttle test (MST) and to determine whether the test needs to be performed more than once to assess the exercise capacity of children and adolescents with cystic fibrosis (CF).

Methods: This was a longitudinal study including patients diagnosed with CF aged more than 6 years. The participants were followed for a period of 9 months and were evaluated at three different time points (visits 1, 2, and 3). Spirometric, anthropometric, clinical, and genetic data were collected, and two MSTs were performed at each visit.

Results: Forty-eight clinically stable volunteers with a mean age of 10.1 ± 2.7 years were initially included. The reproducibility of the test was evaluated using the distance achieved (DA) as the main variable. There were no significant differences in the DA (visit 1, $p = .23$; visit 2, $p = .24$; visit 3, $p = .85$), baseline heart rate (HR) (visit 1, $p = .35$; visit 2, $p = .20$; visit 3, $p = .98$), and peak HR (visit 1, $p = .16$; visit 2, $p = .94$; visit 3, $p = .23$) between the tests performed at each visit. The test-retest reliability demonstrated a high intraclass correlation coefficient at all visits (visit 1, 2, and 3: 0.83, 0.90, and 0.80, respectively) and the variation in HR was the main factor associated with the DA in the MST over time.

Conclusion: The MST was found to be a reproducible and reliable test. The data presented here support the use of a single MST to evaluate and monitor exercise capacity of patients with CF during clinic visits.

KEYWORDS
cystic fibrosis, exercise tolerance, field tests, test reproducibility

1 | INTRODUCTION

Cystic fibrosis (CF) is one of the most commonly inherited genetic diseases causing progressive abnormalities, including complications in the respiratory system, peripheral muscle physiology, and exercise capacity.[1,2] However, CF patients with high levels of physical activity show improved lung function, low hospitalization rates, and high participation in daily living activities.[3–5] In addition, recent evidence shows that impairment of peripheral muscle function is an important systemic disorder caused by CF.[6] Therefore, the evaluation of exercise capacity is considered useful for obtaining information on disease prognosis.[7] It can also be used to develop exercise programs, monitor the effect of interventions, and measure the impact of disease on functional capacity and daily living activities.[2–8]

The cardiopulmonary exercise test (CPET) is considered the gold standard for assessing exercise capacity.[9] However, the specific equipment to perform the test is expensive, restricting its general use in the clinical follow-up of patients with CF.[10] In this context, field tests emerged as an alternative to assess exercise capacity, considering that they are simple and easy to apply in clinical settings.[10,11] The 6-min walk test (6MWT) is one of the most frequently used field tests; although it uses a submaximal protocol,[12] which may not be suitable for individuals with mild lung disease.

On the other hand, the modified shuttle test (MST) is considered a valid alternative for assessing exercise capacity in patients with CF,[2,13] as it is an externally paced, incremental, and maximal exercise test, with a protocol similar to that of CPET.[14,15] However, some studies have suggested the need to perform at least two tests in the first assessment, considering the possible learning effects to performance the test demonstrated when evaluating patients with several diseases,[14,16,17] and no study to date has assessed the need of using multiple tests for children and adolescents with CF.

Considering the complex dynamics of multiprofessional care in specialized CF centers,[18] performing two MSTs at one clinic visit is often considered unrealistic. Therefore, the main purpose of the present study was to assess the need of performing more than one MST to evaluate the exercise capacity of children and adolescents with CF. The subjects were followed during three consecutive clinic visits to a specialized CF center, and the results of two tests conducted on the same day and on different visits were compared; test reliability was also assessed. We hypothesized that, considering the main characteristics of the test protocol, a single test would be sufficient and reliable to evaluate the exercise capacity of children and adolescents with CF.

2 | METHODS

This was a prospective, longitudinal study that evaluated the reproducibility of the MST over a period of 9 months of clinical follow-up of children and adolescents with CF. The study was approved by the Research Ethics Committee under registration numbers 54142716.8.3001.5119 and 1.753013. Patients and their legal guardians were invited to participate in the study on the day of the follow-up visit at the CF specialized center. After the agreement, the consent form was signed, and the individuals included in the study protocol.

The sample size estimate was based on reliability studies with three repetitions per individual, using a minimum acceptable intraclass correlation coefficient (ICC) of 0.60 and an ICC of expected reliability of 0.80. Therefore, a sample size of 33 subjects would provide a power of 80%, with a significance level of 0.05.

Patients of both sexes, aged between 6 and 18 years, under regular follow-up in a rare disease unit of a reference hospital participated voluntarily. The exercise capacity was assessed by the MST, and lung function was evaluated by spirometry. Both evaluations were performed according to the guidelines of the American Thoracic Society and European Respiratory Society (ATS/ERS).[19,20] Anthropometric, clinical, and genetic data were also collected, including the type of mutation and chronic colonization by *Pseudomonas aeruginosa*, which was defined as the persistent presence of the bacterium in the oropharynx swab or sputum culture for at least 6 months or in three consecutive samples.[21] Mutations were classified into three categories: F508del homozygote, F508del heterozygote, and other mutations.

The evaluations were performed at each multiprofessional clinic visit every 3 months. Each volunteer was followed for a period of 9 months, resulting in three clinic visits (visits 1, 2, and 3). At visit 1, two MSTs were performed. Three months later, at visit 2, the volunteers were reassessed using the same tests, and at visit 3, 6 months after the first visit, the last set of evaluations was performed. Only clinically stable children and adolescents were included in the study; stable status was determined according to the assessment of clinical signs of disease exacerbation.[22] Volunteers with pulmonary exacerbation and musculoskeletal, neurological, and hematological alterations that could interfere with the test results were excluded.

2.1 | Anthropometric data

Height was measured using a stadiometer (Caumaq LTDA) with an accuracy of 1 cm; volunteers were instructed to remove their shoes and position themselves with their feet parallel and arms at the sides of the body. Weight measurements were performed in an orthostatic position with minimal clothing, using a balance (IPD7000 Standard) previously calibrated to 50 g. The body mass index (BMI) was calculated using the weight and height (kg/m^2) values, and the WHO Anthroplus program was used to calculate the Z-scores.

2.2 | Exacerbation of clinical signs

To assess the presence of exacerbation, the criterion of Fuchs et al.[22] was used. The presence of four or more clinical signs from among 12 possible ones was indicative of exacerbation.[22]

2.3 | Lung function

Lung function was assessed according to the ATS/ERS recommendations.[20] The spirometric variables analyzed were forced expiratory volume in the first second (FEV_1), forced vital capacity (FVC), forced expiratory flow between 25% and 75% of vital capacity ($FEF_{25\%-75\%}$), and the FEV_1/FVC ratio. They were obtained from the maximum expiratory flow-volume curves based on the values recorded by a Jaeger FlowPro spirometer (Erich Jaeger GmbH). The results were presented as percentages of values calculated using an international reference equation.[23]

2.4 | Modified shuttle test

The MST was performed using the 15-level shuttle test protocol, as described by Bradley et al.[24] The test was carried out in a 10-m corridor, delimited by cones, which had to be bypassed by the participants. The initial walking speed was 0.5 m/s, with increments of 0.17 m/s at each level change, indicated by a triple beep. Each participant underwent the test twice, with an interval of at least 30 min between each test or when the vital signs after the first test had returned to baseline values. Oxygen was offered if required.[25] Both tests were performed by the same physiotherapist. The participants were instructed to walk or run and maintain the speed by following the sound signals; standardized phrases were used at each level change to encourage them.[24] The test was stopped if pale appearance or oxygen saturation below 85% was observed or if the participant was unable to complete the course twice in a row, according to the rhythm of the audio signals. Peripheral oxygen saturation (SpO_2) (INC Model 2500 A Pulse Oximeter; Nonim Medical) and heart rate (HR) (POLAR ElectroOy, Model 90440) were constantly monitored. Respiratory rate, systolic blood pressure, diastolic blood pressure (DBP), and subjective effort (Borg scale)[26] were assessed before and after each test. The distance achieved (DA) was expressed in meters and normalized using reference values.[11] The maximum oxygen consumption (VO_2) was estimated using a previously published equation.[27]

2.5 | Statistical analysis

The quantitative variables were presented as mean and *SD*. The Student *t* test for paired samples was used to compare the data (DA and HR) observed during the tests performed on the same day. For comparison of data recorded over time, data from the best test (greater of the two DA values) was used and analysis of variance for repeated measures was performed, followed by the Bonferroni posttest. For the MST reliability, using the DA as the main variable, the ICC was used, according to the following classification: excellent (ICC ≥0.90), high (0.90 > ICC ≥ 0.70), moderate (0.70 > ICC ≥ 0.40), and low (ICC <0.40).[28] The Bland-Altman analysis was used to identify the accuracy and agreement between two tests performed on the same visit. Statistical significance was set at an alpha level of $p \leq .05$. Finally, preliminary regression models Generalized Estimating Equations - GEE were used to evaluate longitudinally the factors associated with the variation of the DA in the MST over time. Only variables with a *p* value of ≤.05 remained in the final adjustment. The data were analyzed using SPSS version 22.0 (IBM).

3 | RESULTS

A total of 48 children and adolescents (60.4% male) with CF, mean age of 10.1 ± 2.7 years, were included at visit 1. After 3 months, at visit 2, 44 were evaluated, as four participants were excluded because of exacerbation. At visit 3, nine more patients were excluded, and a total of 35 participants were evaluated. A flowchart of patient selection is presented in Figure 1.

Regarding the characteristics of the sample, 54% of the participants showed the F508del heterozygotic genetic mutation, and 16.5% showed chronic colonization by *P. aeruginosa*. The mean percentage predicted FEV_1 (%FEV_1) was 73.5 ± 21.9, and the mean BMI (Z-score) was −1.26 ± 5.3. The characteristics of the samples are presented in Table 1.

When comparing the two tests carried out at the same visit, no significant differences were found in baseline HR (visit 1: $p = .35$; visit 2: $p = .20$; visit 3: $p = .98$), peak HR (visit 1: $p = .16$; visit 2: $p = .94$; visit 3: $p = .23$), and DA (visit 1: $p = .23$; visit 2: $p = .24$; visit 3: $p = .85$) (Figure 2). The Bland-Altman plot shows the mean ± *SD* with a 95% confidence interval ($CI_{95\%}$) of the difference in the DA between test one (T1) and test two (T2) from each visit. From visit 1; 23 ± 134 m ($CI_{95\%}$: −16–62 m), visit

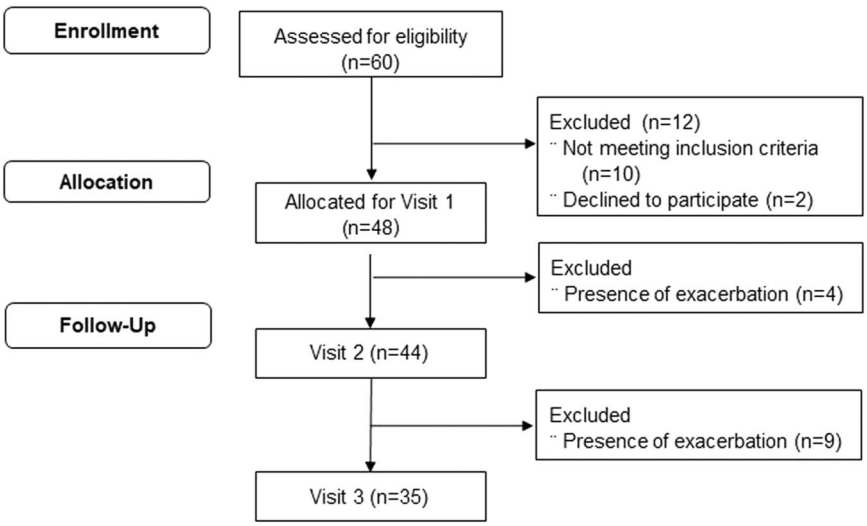

FIGURE 1 Flowchart of the study

TABLE 1 Characteristics of the study sample

Variables evaluated	*n* = 48
Demographic	
Age (years)	10.1 ± 2.7
Male, *n* (%)	29 (60.4)
Anthropometric	
Height (cm)	139.7 ± 15.1
Weight (kg)	32.9 ± 10.1
BMI (Kg/m^2)	16.7 ± 3.5
BMI (Z-score)	−1.26 ± 5.3
Genotyping	
F508del Homozygous, *n* (%)	15 (30.6)
F508del Heterozygous, *n* (%)	26 (54.1)
Other mutations, *n* (%)	7 (14.5)
Chronic airway colonization	
Pseudomonas aeruginosa, *n* (%)	8 (16.6)
Lung function	
FEV_1 (L)	1.6 ± 0.6
FEV_1 (% predicted)	73.5 ± 21.9
FVC (L)	2.0 ± 0.7
FVC (% predicted)	84.2 ± 20.9
FEV_1/FVC (absolute)	79.78 ± 6.95
FEV_1/FVC (% predicted)	84.2 ± 21.0
$FEF_{25\%-75\%}$ (L/min)	1.5 ± 0.8
$FEF_{25\%-75\%}$ (% predicted)	59.5 ± 31.5

Note: Values expressed as mean ± *SD*.

Abbreviations: BMI, body mass index; $FEF_{25\%-75\%}$: forced expiratory flow between 25 and 75% of vital capacity; FEV_1, forced expiratory volume in one second; FVC, forced vital capacity.

2; −18.7 ± 102 m ($CI_{95\%}$: −41–13 m), and visit 3; 10 ± 145 m ($CI_{95\%}$: −39–60 m). No differences (meters) were found between T1 and T2 on visit 1 (23 ± 134; $CI_{95\%}$: −16–62; $p = .24$), visit 2 (−18.7 ± 102; $CI_{95\%}$: −41–13; $p = .23$), and visit 3 (10 ± 145; $CI_{95\%}$: −39–60; $p = .67$) (Figure 3).

When comparing the best physiological responses to the test at each visit, no significant differences were observed, except for the Borg Scale ($p = .05$) and DBP ($p = .01$), but the differences did not seem to be clinically relevant (Table 2).

The test-retest reliability of the MST was evaluated using the DA at each test. The ICC was calculated for each visit, and high coefficients were obtained for all visits (3, 6, and 9 months: 0.83, 0.90, and 0.80, respectively), indicating good reliability. Similar results were observed when the ICC for the best DA at each visit was calculated (0.84) (Table 3).

In the multivariate analysis, the DA ($p = .065$) and the variation in HR ($p = .320$) did not change over time. In the final regression model, only the variation in HR was associated with distanced changes in the MST. The increase of one unit in the variation of the HR causes an increase, on average, of 4.5 m in the DA in the MST (95% CI: 3.4–5.7) $p < .001$.

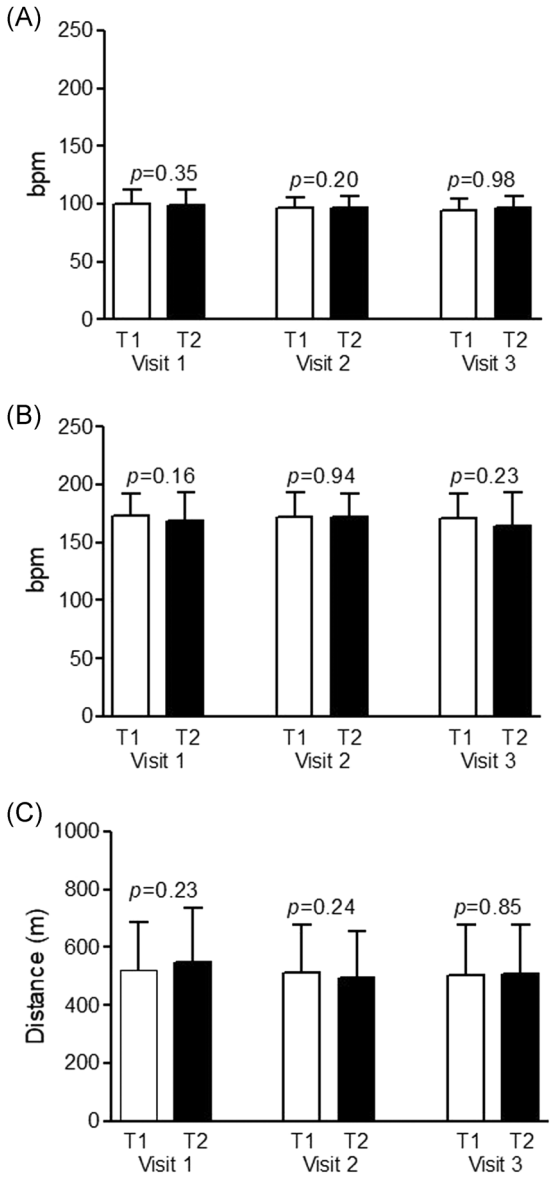

FIGURE 2 Comparisons between two modified shuttle tests performed in the same clinic visit. (A) Resting heart rate. (B) Peak heart rate. (C) Distance achieved. The Student *t* test for paired samples was used. Bpm, beats per minute; T1, test 1; T2, test 2

4 | DISCUSSION

The main findings of the present study confirm the hypothesis that it is not necessary to perform the MST more than once in the evaluation of the exercise capacity of children and adolescents with CF. There were no statistically or clinically relevant differences in the DA values obtained at the two tests performed at visit 1, indicating good test-retest reliability of the MST. This finding is of clinical interest, considering the complex dynamics of multidisciplinary follow-up visits to specialized centers for patients with CF.

The study by Coelho et al.[8] on the accuracy of MST performed in children and adolescents with CF and healthy volunteers aged between 7 and 15 years concluded that a second test is important, as

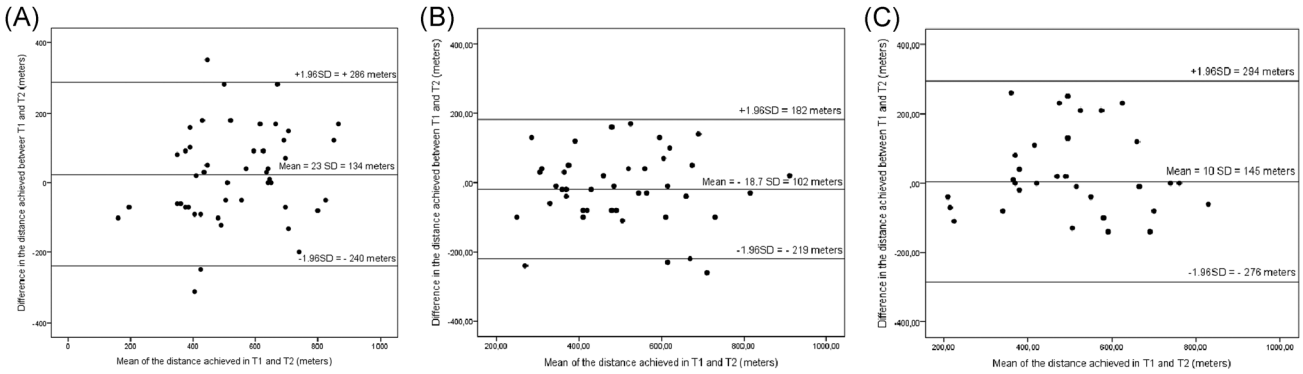

FIGURE 3 Bland-Altman plot showing the individual differences between the two tests (T1 and T2), versus mean values of distance achieved. (A) Visit 1; (B) visit 2; (C) visit 3. The central line indicates the mean difference between paired measurements and the above/below lines indicate upper limit and lower limit with the 95% limits of agreement.

healthy participants experienced difficulties in learning the test and covered a greater distance in the second MST. A difference of 48 m was observed in the DA values between the two tests.[8] However, the group with CF did not have the same difficulties in understanding the MST, as the difference in their DA values was only 16 m (non-significant). In the present study, the participants had no difficulties in performing the test, and no significant differences in the distance covered were found over time to justify the performance of another test. These findings are in accordance with the results of Bradley et al.[24] who evaluated adults with CF who were already familiar with the MST at two visits with an interval of 4–14 days. They concluded that when evaluating exercise capacity, a single MST is sufficient for obtaining precise results, since there were no pronounced variations in the DA values between the two visits.

In the study by Corral et al.,[29] involving children and adolescents with CF, two MSTs were performed at two visits with an interval of 4 days; they concluded that a difference of 97.08 m between the two DA values recorded after a specific intervention period[29] indicated a

TABLE 2 Physiological responses to the modified shuttle test

Variables evaluated	Visit 1 (n = 48)	Visit 2 (n = 44)	Visit 3 (n = 35)	p Value
Baseline				
HR (beats/min)	98.0 ± 12.8	96.3 ± 10	94.8 ± 10,8	.42
SBP (mmHg)	93.7 ± 9,4	92.5 ± 8.6	96.0 ± 9.7	.21
DBP (mmHg)	60.0 ± 5.0	58.1 ± 6.8	58.3 ± 6.4	.30
SpO_2 (%)	96.3 ± 2.3	96.2 ± 2.2	96.0 ± 2.0	.71
RR (rpm)	23.0 ± 6.5	21.7 ± 4.6	20.5 ± 5.7	.12
Borg	0.1 ± 0.4*	0.5 ± 0.8*	0.5 ± 0.8	.05*
Peak exercise				
MST level	8.1 ± 1.5	7.6 ± 1.5	7.7 ± 1.6	.33
MST distance (m)	588.5 ± 168.4	543.4 ± 161.8	558.5 ± 165.3	.41
MST distance (% predicted)	58.25 ± 18.25	52.85 ± 18.73	55.30 ± 21.36	.96
VO_2 estimated (ml/kg/min)	31.48 ± 3.0	30.62 ± 3.0	30.64 ± 3.0	.33
HR (beats/min)	175.9 ± 18.9	174.6 ± 18.6	175.2 ± 18.3	.94
VHR (beats/min)	76.6 ± 24.3	75.6 ± 21.08	80.3 ± 20.1	.64
SBP (mmHg)	111.0 ± 10.7	109.0 ± 12.7	112.1 ± 12.3	.50
DBP (mmHg)	66.0 ± 7.0*	61.5 ± 6.4*	64.0 ± 8.23	.01*
SpO_2 (%)	92.7 ± 4.7	92.8 ± 4.9	91.8 ± 5.1	.59
RR (rpm)	37.0 ± 6.9	37.3 ± 7.9	51.8 ± 73.1	.23
Borg	6.1 ± 2.5	5.8 ± 2.6	6.2 ± 2.2	.77

Note: Values expressed as mean ± *SD*. One-way ANOVA followed by the Bonferroni posttest.
*Statistical significance set at $p < .05$.

Abbreviations: ANOVA, analysis of variance; DBP, diastolic blood pressure; HR, heart rate; MST, modified shuttle test; rpm, respirations per minute; RR, respiratory rate; SBP, systolic blood pressure; SpO_2, peripheral oxygen saturation; VHR: variation of heart rate (HR peak exercise − HR baseline); VO_2, oxygen consumption.

TABLE 3 Reliability of the distance achieved in the modified shuttle test (MST) in each clinic visit and between different visits

	Intraclass correlation coefficient (ICC)*
Same visit	
Visit 1: T1 × T2	0.83 (0.70–0.90)
Visit 2: T1 × T2	0.90 (0.80–0.94)
Visit 3: T1 × T2	0.80 (0.60–0.90)
Between visits (best test of each visit)	
Visit 1 × visit 2 × visit 3	0.84 (0.70–0.90)

Note: *Values expressed as ICC (95% confidence interval). T1: test 1; T2: test 2.

clinically significant difference. According to the ATS/ERS,[19] the minimum distance indicating clinical improvement after an intervention in adult patients is 47.5 m in the incremental shuttle test (ISWT). In the present study, the highest variation in the DA was 45 m, and age did not affect distance variability. We assessed patients with CF using three distinct evaluations (clinic visits) with a minimum interval of 3 months between them. Although we speculated that the 3-month interval between tests could be an important factor in reducing reproducibility, which would reinforce the requirement of two tests in each evaluation,[30,31] our results strongly refuted this assumption, as the longitudinal follow-up of patients with stable CF disease showed that one MST is sufficient to assess the patient's exercise capacity.

HR response is an important variable of the MST that indicates the cardiopulmonary performance of patients with CF.[2] In the present study, no significant differences were found in the HR among the three different evaluations, demonstrating that the cardiac overload was similar in the test-retest over time. Lans et al.[32] assessed adults with heart failure using the 6MWT at 3-, 6-, 9-, and 12-month follow-up visits and evaluated the HR responses over time and the findings of these authors were similar to those of the present study. For these patients, the long-term response to exercise does not require two tests, as cardiac overload did not vary significantly between tests. Bradley et al.[24] found no significant differences in the HR at rest and HR at peak exercise in adults with CF. The findings of these authors are similar to ours and confirm that the long-term evaluation of the response to exercise does not require the performance of two tests as the cardiac overload did not vary significantly between the evaluations.

CPET is carried out frequently to assess exercise tolerance in patients with chronic lung disease, as its results are directly associated with morbidity and mortality according to functional impairment.[7,33] However, considering the complexity of the evaluation and the available resources, this tool is still not regularly used in specialized CF centers.[10,33] Similar to many CPET protocols, MST uses an incremental maximum protocol[33,34] that is externally controlled by an audio signal, making it possible to estimate the maximum oxygen consumption (VO_{2max}).[25,27] Indeed, the test has already been validated for adults,[34] adolescents, and chidren,[8] making it a good alternative to clinical field tests, and it is reproducible in the assessment of children and adolescents with mild respiratory disease.[35] The systematic review by Parreira et al.[36] indicates that MST is a reliable test for the evaluation of chronic respiratory diseases. It is reproducible in healthy children[37] and in children with CF but not in children with significant pulmonary impairment.[7,25,30–38] The findings of the present study confirm the good reproducibility of the MST in children and adolescents with mild pulmonary impairment. We believe that the sound signal used during the MST is a key factor contributing to the high reliability, as the external beep controls the participant's rhythm.[25] The sound enables a controlled increase in speed,[14] which minimizes the rhythm change during the test, regardless of the participants' motivation, and may help reduce the learning effects, as demonstrated in the present study.

The reliability of the MST in children and adolescents with CF is still poorly described, although it is well documented in the adult population with CF.[24,34–39] The present results demonstrated that the MST has high test-retest reliability and high reproducibility between tests applied over time. These results are similar to the findings of Corral et al.[29] in a sample of children and adolescents with CF, although the interval between tests was shorter. This may have contributed to the maintenance of test performance and the good reliability. Studies have shown that HR behavior is an important indicator of functional performance in the MST.[2,21] Reports have shown that the increase in resting HR is associated with a reduction in the DA in patients with CF.[30] In the present study, when the determinants of the DA over time were evaluated, this association was also observed. The variation in HR assessed by the difference between peak and baseline HR may be considered as an indicator of overload with direct repercussions on the DA in the MST.

The present study also has limitations, including the absence of gas analysis, which could precisely detect exercise intolerance. However, as a field test, the MST is intended to be an alternative in clinical practice whenever the gold standard CPET is not possible or available, allowing the estimation of exercise capacity through the DA and the evaluation of the cardiopulmonary performance through the chronotropic response. The fact that the study sample consisted of patients with mild pulmonary impairment may have also influenced the results, as the exercise response may vary according to disease severity.

In conclusion, our results indicate that a single MST is sufficient in the evaluation of children and adolescents with CF. MST is considered a reproducible and reliable test for this population. The cardiovascular variables and DA are the most appropriate test variables for monitoring the performance of this population over time.

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AUTHOR CONTRIBUTIONS

Luanna Rodrigues Leite: conceptualization (equal); data curation (equal); formal analysis (equal); funding acquisition (equal); investigation (equal); visualization (equal); writing original draft (equal); writing review & editing (equal). **Karen Caroline Vasconcelos Queiroz**: conceptualization (equal); data curation (equal); funding acquisition (equal); investigation (equal); methodology (equal); project administration (equal); writing original draft (equal); writing review & editing (equal). **Franciely Helena da Silva**: data curation (equal); formal analysis (equal); investigation (equal); methodology (equal); writing original draft (equal). **Cristiane Cenachi Coelho**: conceptualization (equal); formal analysis (equal); investigation (equal); methodology (equal); project administration (equal); supervision (equal); validation (equal); visualization (equal); writing original draft (equal); writing review & editing (equal). **Márcio Vinícius Fagundes Donadio**: data curation (equal); formal analysis (equal); funding acquisition (equal); investigation (equal); methodology (equal); resources (equal); validation (equal); visualization (equal); writing original draft (equal); writing review & editing (equal).

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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