ABSTRACT: Preschool-age children with neuromuscular disorders are often excluded from clinical trials due to the lack of reliable and objective strength measures. We evaluated the reliability of measuring foot and ankle muscle strength in 60 healthy children age 2-4 years. The strength of foot inversion and eversion, ankle plantarflexion, and dorsiflexion was measured using a hand-held dynamometer. Intrarater and interrater reliability of two assessors was determined for each muscle group using intraclass correlation coefficients (ICCs), 95% confidence intervals (CIs), and standard error of measurement (SEM). For all muscle groups intrarater (ICC_{2,2} = 0.85-0.94, 95% CI = 0.75-0.96, SEM = 1.5-4.7 N) and interrater (ICC_{2,1} = 0.88-0.96, 95% CI = 0.81-0.98, SEM = 1.2-4.6 N) reliability was high for all children. As expected, reliability was generally highest in 3- and 4-yearold children and lowest in 2-year-old children. Hand-held dynamometry can reliably measure foot and ankle strength in very young children and may help aid in diagnosis and in characterizing disease progression in disorders affecting the foot and ankle.

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RELIABILITY OF QUANTIFYING FOOT AND ANKLE MUSCLE STRENGTH IN VERY YOUNG CHILDREN

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There are few reliable or objective measures of foot and ankle muscle strength for children under the age of 4 years. Such measures are required for clinical trials examining interventions to improve foot and ankle muscle weakness in children with neuromuscular disorders and will become essential over the coming years with therapies emerging for peripheral neuropathies such as Charcot–Marie–Tooth disease. Reliable and objective measures of foot and ankle strength are also required to obtain normative reference values, which may assist in detecting weakness in young children presenting with possible neuromuscular disorders, as well as in other

patient populations such as juvenile arthritis, cerebral palsy and connective tissue disorders.

Muscle strength is traditionally measured by manual muscle testing using the Medical Research Council (MRC) grades.²² This method is inexpensive and quick to conduct, but has limited sensitivity, particularly in those with mild weakness.11 Even when implemented by experienced evaluators, manual muscle testing has lower reliability and accuracy than other more objective and quantitative instrumented strength evaluation.^{9,18} One of these instruments, the hand-held dynamometer, is a portable digital device incorporating a calibrated load cell. In contrast to fixed dynamometry such as the Kin-Com system (Chattecx, Chattanooga, Tennessee), the hand-held dynamometer is commonly used in the clinical setting to measure isometric muscle strength because it is lightweight, inexpensive, and easy to use.30 Isometric foot and ankle strength can be measured by hand-held dynamometry with high intrarater and interrater reliability in both healthy adolescents and adults^{5,30,32} as well as in adolescents and adults with neuromuscular disorders such as spinal muscular atrophy and Charcot-Marie-Tooth

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Abbreviations: CI, confidence interval; HHD, hand-held dynamometer; ICC, intraclass correlation coefficient; VAS, visual analog scale

Key words: children; hand-held dynamometry; manual muscle testing; quantitative muscle testing; reliability; strength

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disease.5,20 Reliable strength measures have also been obtained using hand-held dynamometry in healthy children older than 5 years^{7,14} and in school-aged children with neuromuscular disorders such as Duchenne muscular dystrophy and spinal muscular atrophy. 18,20,28 However, very few studies have investigated the reliability of hand-held dynamometry in preschool children, i.e., under the age of 4 years, who are often excluded from reliability studies and clinical trials because of their perceived inability to understand and comply with strength testing procedures.3,4,28 Furthermore, normative reference values have not been reported for many muscle groups in children under the age of 4 years,1 and those that are reported have tended to group children in a 3-5-year age band, which is inappropriate given the rapid growth and development that occurs during this period.

Only one study has evaluated the reliability of handheld dynamometry to measure muscle strength in children under the age of 4 years. Gajdosik¹² assessed intrarater reliability of measuring isometric strength of the shoulder, elbow, and knee flexors/extensors in 45 children with typical development age 2, 3, and 4 years and found high reliability coefficients for the muscle groups in all children [intraclass correlation coefficient (ICC)_{2.1} 0.90-0.95]. This was irrespective of challenging behaviors such as inattention, resistance and fussiness. However, foot and ankle strength was not investigated and interrater reliability was not assessed. Therefore, the aim of our study was to evaluate the intrarater and interrater reliability of measuring foot and ankle muscle strength in 60 healthy children aged 2-4 years with hand-held dynamometry.

MATERIALS AND METHODS

Participants. A community sample of 60 children aged 2–4 years with normal development participated in this study. Each yearly age group comprised 20 children (10 boys and 10 girls). The children were recruited through advertisements in the hospital media and through local community child care centers. Children were excluded if they were born prematurely (<37 weeks gestation), failed to demonstrate normal age-related development according to the well-validated Ages and Stages Questionnaire, ^{17,25} or had a family history of a medical condition affecting foot and ankle muscle strength. The study was approved by the Human Research and Ethics Committee of the Children's Hospital at Westmead.

Instrument. Isometric muscle strength was quantified using the Citec hand-held dynamometer (CIT

Technics, Groningen, The Netherlands). The handheld dynamometer was calibrated as per the manufacturer's specification, to an error margin of 0.1% and a range of 0 to 500 N. The hand-held dynamometer was esthetically modified in order to appear less frightening and more acceptable to the young participants through the application of age-appropriate decorations.

Procedure. The children were assessed independently by two assessors, assessor 1 (K.R.), a physical therapist with 7 years of clinical pediatric experience, and assessor 2 (J.B.), a podiatric physician with 10 years of clinical experience. Each child was positioned in long sitting (hips flexed and knees extended) on an examination table with a backrest, and isometric muscle strength of foot inversion and eversion, and ankle plantarflexion and dorsiflexion was measured according to a standardized procedure. The seated position was used for all movements so the children could visualize the procedure to minimize interruptions and positional changes between testing of the various muscle groups.^{8,12} The assessor stabilized the lower limb proximal to the ankle joint to isolate movement at the joint and minimize additional and substitution movements. For inversion, the dynamometer was positioned against the medial border of the foot, just distal to the base of the first metatarsal. For eversion, the dynamometer was placed against the lateral border of the foot, just distal to the base of the fifth metatarsal; for ankle plantarflexion against the plantar surface of the foot, just proximal to the metatarsal heads; and for ankle dorsiflexion, against the dorsal surface of the foot, just proximal to the metatarsal heads. For all muscle contractions, the ankle joint (talocrural) and subtalar joint (talocalcaneal) was positioned in mid-range of the overall length of the muscle in accordance with the optimal test position for two or more joint muscles.¹⁵ No part of the foot was touching the testing surface during the procedure and the heels rested over the edge of the examination table.

The 'make' test method was used whereby the assessor holds the hand-held dynamometer stationary while the child exerts a maximal force against it. Previous studies have reported the highest reliability with the make test, as opposed to the 'break' test, whereby the assessor attempts to overcome the maximal effort of the child.²⁷ Each movement was explained, demonstrated, and practiced with each child in a manner appropriate to performance of the age until the assessor felt the movement was correct and to the best of the child's ability.

Three consecutive contractions of 3–5 s for each muscle group were measured by both assessors (randomly ordered) and recorded by a research assistant to reduce measurement bias. The average of three contractions (one trial) was used for analysis since mean values are considered more reliable than maximal values.31 Standardized verbal encouragement was given to each child with every effort. Short rest breaks were given between trials to allow the child to recover from previous efforts and to enable comforting by a parent or guardian. Further contractions were carried out if the assessor felt the child's effort was incomplete, or the movement was performed incorrectly. A modified 100-mm visual analog scale (VAS) was used to record each assessor's perception of participant behavior during testing.²¹ A measure of 0 was indicative of very challenging behavior and of 100 was indicative of excellent behavior.

To determine intrarater and interrater reliability, the testing procedure was performed twice by each assessor. Assessor 2 left the room while assessor 1 performed strength testing and vice versa. The second session of testing was conducted later on the same day. One foot only from each child was randomly selected for testing, to satisfy the independence requirement for statistical analysis, ¹⁹ and to minimize any bias that may have originated from assessor (and participant) limb dominance, fatigue, improved skill acquisition, or any other unknown cause of systematic error.

Statistical Methods. Descriptive statistics were calculated to characterize the study sample in SPSS v. 15.0 (Chicago, Illinois). ICCs and 95% confidence intervals (CIs) were calculated to determine intrarater reliability (ICC_{2.1}) and interrater reliability (ICC_{2.2}).²⁴ Benchmarks suggested by Fleiss¹⁰ were used to interpret ICC values, where a value of 0.75 or greater indicates excellent reliability; 0.40 to 0.75, fair to good reliability; and 0.40 or less, poor reliability. To determine the absolute between-trial variability in scores, the standard error of measurement (SEM) was calculated.²⁴ Strength measurements are presented as raw strength scores for individual muscle groups, as well as strength scores normalized to body weight (expressed as a percentage of weight) since body weight correlates strongly with strength in children and is considered more clinically meaningful than raw scores.^{2,26}

RESULTS

Strength values were obtained from 60 children aged 2, 3, and 4 years. Physical characteristics of the three groups of children were: 2-year-olds (mean age

28.7, SD 3.0, range 24–34 months; mean height 90.0, SD 3.3, range 85.0–97.0 cm; mean weight 13.6, SD 1.7, range 11.2–16.5 kg); 3-year-olds (mean age 40.1, SD 3.2, range 36–47 months; mean height 99.4, SD 3.0, range 70.0–106.1 cm; mean weight 16.2, SD 1.4, range 12.8–18.0 kg); and 4-year-olds (mean age 53.9, SD 0.11, range 48–59 months; mean height 108.4, SD 4.8, range 101.4–118.2 cm; mean weight 18.2, SD 2.7, range 26.8–18.2 kg). As expected, there were significant differences between children of different ages for height (F = 115.437, P < 0.001) and body weight (F = 26.225, P < 0.001). There were no significant differences between boys and girls for age (t = -0.035, P = 0.972), height (t = 0.680, P = 0.499), or body weight (t = 1.709, P = 0.093).

Intrarater Reliability. For all children the ICCs and 95% CIs for assessor 1 were excellent (ICC $_{2,1}=0.90-0.94$, 95% CI = 0.83-0.97) and the measurement error was low (SEM, 2.15-4.59 N). For assessor 2 the ICCs and 95% CIs were also excellent for all children (ICC $_{2,1}=0.88-0.95$, 95% CI = 0.81-0.98) and measurement error was low (SEM, 1.25-2.80 N). For both assessors, ankle dorsiflexion was least reliable (ICC $_{2,1}=0.88-0.90$) and plantarflexion was most reliable (ICC $_{2,1}=0.94-0.96$). Intrarater reliability was generally highest in 4-year-old children (ICC $_{2,1}=0.78-0.96$) followed by children aged 3 years (ICC $_{2,1}=0.79-0.94$) and 2 years (ICC $_{2,1}=0.62-0.91$). Full details of the intrarater reliability are presented in Supplementary Table 1.

Interrater Reliability. For all children the ICCs and 95% CI were excellent (ICC_{2,2} = 0.85–0.94, 95% CI = 0.75–0.96) for all muscle groups and the measurement error was small (SEM = 1.54–4.67 N), indicating that measurements varied little between trials. Again, ankle dorsiflexion (ICC_{2,2} = 0.85) was the least reliable and plantarflexion was the most reliable (ICC_{2,2} = 0.94). Interrater reliability was highest in 3-year-old children (ICC_{2,2} 0.77–0.93) followed by children aged 4 years (ICC_{2,2} 0.69–0.90) and 2 years (ICC_{2,2} 0.50–0.83). Full details of the interrater reliability are presented in Supplementary Table 2.

Behavior. Types of challenging behaviors included lack of cooperation and attentiveness. As expected, children age 2 years obtained the lowest behavior scores (mean 48.3, SD 27.5 mm) followed by the 3-year-olds (83.8, SD 15.9 mm) and 4-year-old children (93.7, SD 12.6 mm). While these differences were statistically significant (F = 29.307, P < 0.001),

Table 1. Normative strength data for foot inversion and eversion, ankle dorsiflexion, and plantarflexion in healthy children aged 2-4 years.

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Movement	Raw (N)	Normalized (% BW)
Inversion		
All	41.1 (17.7)	26 (9)
2 years	25.8 (8.1)	19 (6)
3 years	42.0 (15.3)	26 (9)
4 years	55.5 (14.5)	31 (7)
Eversion		
All	39.5 (17.7)	25 (9)
2 years	24.2 (8.6)	18 (6)
3 years	39.9 (13.2)	25 (8)
4 years	54.4 (15.4)	31 (9)
Dorsiflexion		
All	44.5 (17.9)	28 (9)
2 years	29.6 (8.9)	18 (6)
3 years	43.9 (45.4)	28 (9)
4 years	60.0 (14.0)	34 (7)
Plantarflexion		
All	117.6 (55.9)	74 (31)
2 years	70.1 (29.8)	53 (24)
3 years	120.6 (45.4)	76 (28)
4 years	162.0 (48.1)	92 (30)

Values are mean (SD). N = 60 (20 per age group including 10 boys and 10 girls).

there were no significant differences between boys and girls.

Normative Reference Values. Table 1 presents normative reference strength data for foot inversion and eversion as well as ankle plantarflexion and dorsiflexion obtained by averaging the four trials collected by both assessors (12 contractions) of all 60 children to ensure a true representation of their strength capabilities and provide a closer estimate of actual strength. Muscle strength was found to increase linearly with age (r = 0.66-0.71, P < 0.001), height (r = 0.70-0.72, P < 0.001), and body weight (r = 0.57-0.68, P < 0.001). Strength did not differ significantly between boys and girls.

DISCUSSION

We have demonstrated acceptable intrarater and interrater reliability of quantifying foot and ankle muscle strength using hand-held dynamometry in very young children. Under controlled testing conditions, high correlation coefficients and low measurement error can be achieved in children age 2–4 years. These results suggest that hand-held dynamometry may be a suitable instrument with which to measure the presence, severity, and progression of disorders affecting foot and ankle strength in preschool-age children. Our findings are in accordance

with previous work establishing hand-held dynamometry as a reliable and valid tool for assessing foot and ankle muscle strength in both healthy and affected school-age children^{13,29,31} and adults,^{5,30} We also confirm the results of a previous reliability study reporting the clinical acceptability of hand-held dynamometry in very young children aged 2–4 years.¹²

Interestingly, although behavior improved with maturation, interrater reliability did not. Muscle strength was most reliable in 3-year-old children followed by the 4- and 2-year-old children. Gajdosik 12 also reported highest reliability for 3-year-old children (ICCs_{2,1} = 0.85–0.92) compared to 4-(ICCs_{2,1} = 0.54–0.94) and 2-year-olds (ICCs_{2,1} = 0.76–0.91) when using hand-held dynamometry to assess the strength of proximal muscle groups. Similarly, the presence of challenging behaviors did not seem to strongly influence reliability. We suggest age and behavior should not be seen as a barrier to reliable strength testing.

Gender had no effect on reliability or overall strength measures. This finding is consistent with previous studies reporting normative reference values for isometric muscle strength in older children using hand-held dynamometry, as it appears that gender differences in strength are not apparent until puberty. 1-3 Therefore, we combined strength values for boys and girls in the establishment of normative reference data (Table 1). We also presented normative strength data both as raw scores and as normalized scores, i.e., strength scores as a percentage of body weight. Since strength correlates with body weight in children, normalized strength scores may provide a more useful representation of the child's strength.^{2,26} For example, clinically it may be more meaningful to report a child was able to exert 60% of their bodyweight for ankle plantarflexion rather than reporting they were able to exert 120 N.

As foot and ankle muscle strength can be measured reliably by hand-held dynamometry in healthy children aged 2-4 years, this technique may be useful to aid diagnosis, characterize disease progression, and as an outcome measure in trials of neuromuscular disorders. Indeed, it has been recently reported that asymptomatic healthy populations produce less reliable results on repeat muscle testing than symptomatic patient populations.⁶ Therefore, dynamometric strength testing could possibly have greater reliability in children with neuromuscular disorders. However, as we have only conducted this study in a healthy cohort of preschool-age children, further disease-specific testing in children with neuromuscular disorders is required to determine the suitability of handheld dynamometry in affected children. Reliability studies involving multiple assessors would also confirm whether this method is suitable for use in multicenter clinical trials to evaluate therapies that improve foot and ankle strength in very young children with neuromuscular disorders.

This study has some limitations. First, each assessor was not blinded to the read-out on the dynamometer during the testing process. While our procedure was optimal, given the specifications of the Citec hand-held dynamometer and the behavioral challenges of children aged 2-4 years, some potential bias was unavoidable. For instance, to activate the hand-held dynamometer an on/reset button is pressed and a small wheel is carefully adjusted to zero before it is ready to measure. If the assessor was blinded to this procedure the dynamometer may not have been appropriately zeroed. Furthermore, there is an auto shutoff after 15 s and blinding of the digital read-out would have resulted in wasted efforts by the participant and potential worsening behavior. To reduce the risk of bias, a research assistant read and recorded all strength values on the dynamometer after each contraction (12 per foot per session for each assessor).

Second, while all our mean ICCs for intrarater and interrater reliability were regarded as good to excellent, 10 some of the 95% CIs were quite wide, particularly for muscle strength in 2-year-old children. In addition, the interrater SEM for ankle plantarflexion and dorsiflexion in 2-year-old children was large, representing \approx 21%–25% of mean strength. This contrasts with 4%–11% for 3- and 4-year-old children. Closer examination of the individual strength data revealed that three children in the 2-year-old group produced highly variable efforts which increased 2–3-fold between trials 1 and 2. This variability may relate to a learning effect, limited motivation, or the fact that 2-year-old children generally produced more challenging behaviors.

Third, while we adopted a standardized protocol, gave the children an abundance of encouragement, and provided clear verbal cues to ensure the children were pushing the dynamometer as hard as they could, the validity of the strength measures was not investigated in this study. The generally high interrater reliability suggests the children were repeatedly giving their best efforts, but validation of the dynamometric measures against more functional tasks requiring high level foot and ankle muscle strength such as running and jumping may help determine whether the strength data obtained are representative of maximal strength. Further research addressing these limitations is required to determine the

efficacy of many promising therapies for pre-school age children with muscle and nerve disorders.

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