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Original Article

Measurement Repeatability and Reproducibility of Virtual Goniometry of a Set of Acquired Images in Youths with Arthrogryposis Multiplex Congenita

Marianne Gagnon[1,2], Gabriela Marino Merlo[1], Noémi Dahan-Oliel[1,3], Louis-Nicolas Veilleux[1,2]

[1]Shriners Hospitals for Children-Canada, Montreal, Canada;
[2]Department of Surgery, Division of Experimental Surgery, McGill University, Montreal, Canada;
[3]Faculty of Medicine and Health Sciences, School of Physical & Occupational Therapy, McGill University, Montreal, Canada

Abstract

Objectives: To evaluate the intra-rater repeatability and the inter-rater reproducibility of using a virtual goniometer to measure upper and lower extremity joint range of motion (ROM) in youths with arthrogryposis multiplex congenita (AMC). **Methods**: Youths presenting with AMC aged 8 to 21 years old were recruited. ROM of the upper and lower limbs were assessed remotely during a teleassessment on a video-conferencing platform. Screen captures were taken and ROM were measured by two raters, two-weeks apart, using a virtual goniometer. Intraclass correlation coefficient (ICC) and associated 95% confidence interval (CI) were calculated to assess intra-and inter-rater repeatability and reproducibility. **Results**: Nine participants were included with a median age of 15.9 years (range: 11.3 to 20.8 years). The overall intra-rater ICC was 0.997 (95% CI:0.996 to 0.997) for the first rater and 0.993 (95% CI:0.992 to 0.994) for the second rater. The inter-rater ICC ranged from 0.410 (95% CI:-0.392; 0.753) for forearm pronation to 0.998 (95% CI:0.996; 0.999) for elbow flexion. **Conclusions**: Results of the current study suggest that virtual goniometry is reproducible and repeatable for the ROM of most joints. Future studies should evaluate procedural reliability and validity of the proposed method for youth with complex conditions.

Keywords: Arthrogryposis Multiplex Congenita, Joint Range of Motion, Teleassessment, Telerehabilitation, Virtual Goniometry Assessment

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Introduction

Arthrogryposis Multiplex Congenita (AMC) is defined as a group of rare musculoskeletal conditions characterized by multiple congenital non-progressive joint contractures and muscle weakness[1]. One of the main clinical problems of youths with AMC is that they have significant ROM limitations that may lead to compensations, pain, activity limitations and participation restrictions[2,3]. Joint contractures occur when full passive range of motion (ROM) is not present, and can affect the limbs, spine, and jaw in individuals with AMC[1]. These contractures can be exacerbated by muscle imbalances or the absence of a functioning muscle[4]. The stronger muscle will pull the joint into a more contracted position as the weaker antagonist muscle is unable to overpower it. It is therefore important to enhance muscle strength and mobility, and to maintain optimal joint position through exercise and stretching. These modalities can help maintain maximum ROM and prevent joint contractures[4-7]. Therefore, joint contractures and associated ROM limitations are the main impairments targeted during rehabilitation for this population. For example, measurement of baseline active and passive ROM using goniometry is essential to determine the need for both surgical and/or conservative orthopedic interventions intended to improve mobility, function and ultimately quality of life[8]. Additionally, ROM measurement is utilized to assess treatment efficacy (e.g. bracing, physical

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Corresponding author: Marianne Gagnon M.Sc., 1003 Decarie Bld, Montréal, Qc, H4A 0A9, Canada
E-mail: marianne.gagnon3@mail.mcgill.ca

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Figure 1. Example of a youth with arthrogryposis multiplex congenita and associated joint contractures and skin webbing.

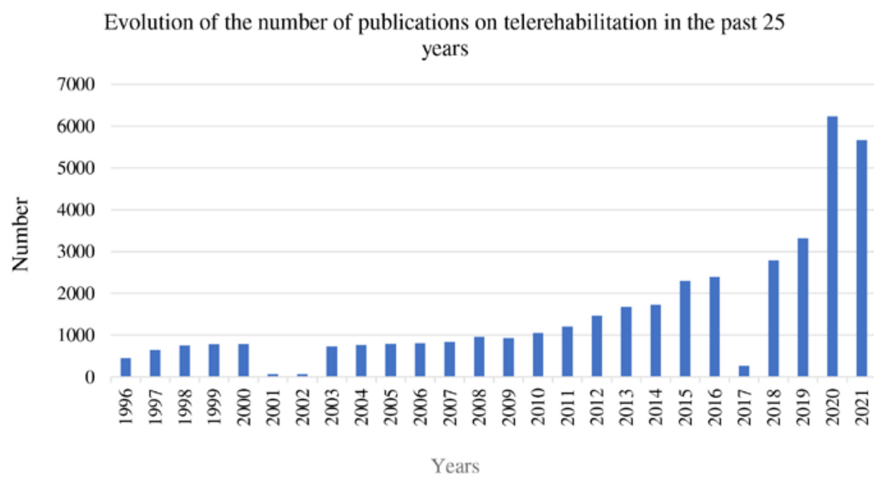

Figure 2. Increased number of publications on telerehabilitation in the past 25 years based on the keyword search "Telemedicine", "Teleassessment", "Telerehabilitation" or "Telehealth" in PubMed (Date accessed: June 13, 2022).

therapy, surgery) in youth with AMC. In addition to limb involvement, AMC may also involve other systems (e.g., gastro-intestinal, genito-urinary)[1]. Therefore, given the rarity and complexity of AMC, specialized multidisciplinary care is required. This specialized care is most often provided in large urban areas, which may not be easily accessible for many individuals with AMC. Consequently, developing approaches allowing for remote management of the patient's clinical problems may benefit this population. However, remote measurement of ROM in youths with AMC can raise additional challenges secondary to particularities such as skin webbing or joint contractures (Figure 1). These particularities make it more difficult to identify the bony landmarks or to perform movements in a single plane of motion in front of a camera.

The technology to perform remote assessment of ROM in an AMC population has recently started to be developed. Telerehabilitation is defined as the use of telecommunication technologies for the purpose of delivering rehabilitation services remotely[9]. The interest in this domain has been gradual since 2015, but a more rapid growth is noticeable between the years 2019-2021 as seen on Figure 2. The Covid-19 worldwide pandemic may be a contributing factor to the increased interest and need for implementation in several digital practice opportunities, such as telerehabilitation[10]. Currently, telerehabilitation is being used to provide remote evaluations and interventions to patients, allowing for greater accessibility, reduced costs, and lesser travelling time, especially for those requiring specialized rehabilitation services[11,12].

In recent years, few studies have assessed the feasibility and reliability of remote assessment of ROM using photographs, screen captures and goniometer applications/software. Studies evaluating measurement reliability, i.e., repeat measurements of the acquired image(s), (as opposed to procedural reliability, where the entire measurement procedure is repeated including patient positioning, image capture, and subsequent measurement), suggests good to excellent reliability[13,14]. Mehta et al. compared measurement reliability assessed remotely and in person and found comparable results[14]. However, in these studies, ROM assessment was limited to ROM in the sagittal plane (i.e., knee flexion and extension, wrist flexion and extension). Measurement reliability for remote assessment of ROM in other planes remains unstudied. Further, prior studies have assessed ROM in healthy, high-functioning individuals and in controlled environments (e.g., clinical settings). Therefore, it remains unclear if the condition of individuals with AMC (e.g., limited ROM and skin webbing), or if measuring ROM virtually in less controlled environments (e.g., at home) impairs measurement reliability. When assessing in-person repeatability and reproducibility of ROM measurement in children with cerebral palsy, a population similar to AMC, Mutlu et al. found good to excellent between-days procedural repeatability and within-day reproducibility for ankle dorsiflexion, which is similar to what was found in children without musculoskeletal conditions[15,16].

The aim of this study is to evaluate the measurement repeatability and reproducibility of using a virtual goniometer for remote active ROM measurements of a set of acquired upper and lower extremity joint images in youths with AMC. This is a sub-study for which the main goal was to prescribe a home-based exercise program where participants performed meaningful tasks and activities to indirectly promote active ROM. Active ROM was therefore the comparison measure that was taken, and it was more feasible to measure remotely, without assistance. The acquired images were used in this subsequent study[12]. Repeatability refers to the variation in repeat measurements made on the same subject under identical conditions while reproducibility refers to the variation in measurements made on a subject under changing conditions (e.g. different raters)[17]. This study focuses specifically on measurement repeatability and reproducibility and does not evaluate procedural reliability. It was hypothesized that the inter-and intra-rater repeatability and reproducibility would be acceptable for clinical use, but less reliable than reported in previous studies due to particularities among youth with AMC and video capture conducted by the family, as opposed to high-tech video equipment with stringent protocols in the clinical settings.

Materials and Methods

The data presented in this study are a subset of a previous telerehabilitation pilot study[12,18]. Individuals were asked to participate if they were between 8 and 21 years of age, had a clinical diagnosis of AMC and understood written and spoken English or French. Exclusion criteria included recent surgery (3 months for soft-tissue, 6 months for bony surgery), cognitive deficits or residency outside of Canada. Youths aged 14-21 years completed an informed consent forms while an assent was provided for those aged 8-13 years. In the context of the main study, all parents completed a consent form.

A remote multidisciplinary teleassessment including active ROM assessment was conducted by a physical therapist and an occupational therapist prior to commencing the telerehabilitation intervention as well as at the end of the 12-week intervention[12]. At the beginning of the session, participants were asked to inform the therapists if pain occurred during the assessment. All teleassessments were performed synchronously using Zoom Pro (Zoom Video Communications Inc.), a video conferencing system. The two above-mentioned clinicians were on-site (e.g., in their offices) while participants were in their regular environments (e.g., home, school). A person (e.g. parent, school therapist) was present and called upon to assist with camera and participant positioning, when necessary. During the ROM assessment, screen captures of joint angles were taken for both upper and lower extremity joints. Details for the protocol of the 12-week intervention are provided elsewhere[12,18].

Two raters (MG, GMM) who have 4 and 5 years of experience as a kinesiologist and physiotherapy technologist, respectively, reviewed existing screen captures from the previous telerehabilitation pilot study. Screen captures demonstrating: 1) extreme physiological compensations accompanying the desired joint movement and, or 2) improper plane of movement, were identified and excluded from measurement and analysis in the present study based on rater-agreement. Before the start of the study, both raters did not have prior experience with the virtual goniometer; therefore, they practiced on three sample participants, excluded from the current study, prior to data collection to ensure consistency in angle measurement. Both raters then proceeded to measure ROM. Existing screen captures were uploaded into Kinovea, version 0.8.15, a freely accessible software that allows for measurement of each ROM with a virtual goniometer. Joint angles for each accepted screen capture were measured by placing the virtual fulcrum and both proximal and distal arms in concordance with standard goniometry procedures (Figure 3)[19]. The main difference with standard measurement (i.e. in person) is that to obtain direct measurement (i.e. without the need of calculation), the arm of the goniometer need to be positioned on the prolonged line of the proximal segment (Figure 3b). This process was completed twice by both raters with at least 2-week interval in between measurement. Both raters were also blinded to one another's results. All measurements were recorded into individual Excel spreadsheets. Data collected by each rater was then merged into a final Excel spreadsheet to allow for statistical analysis. Repeated measures were analyzed for each joint with respect to a within-subjects design.

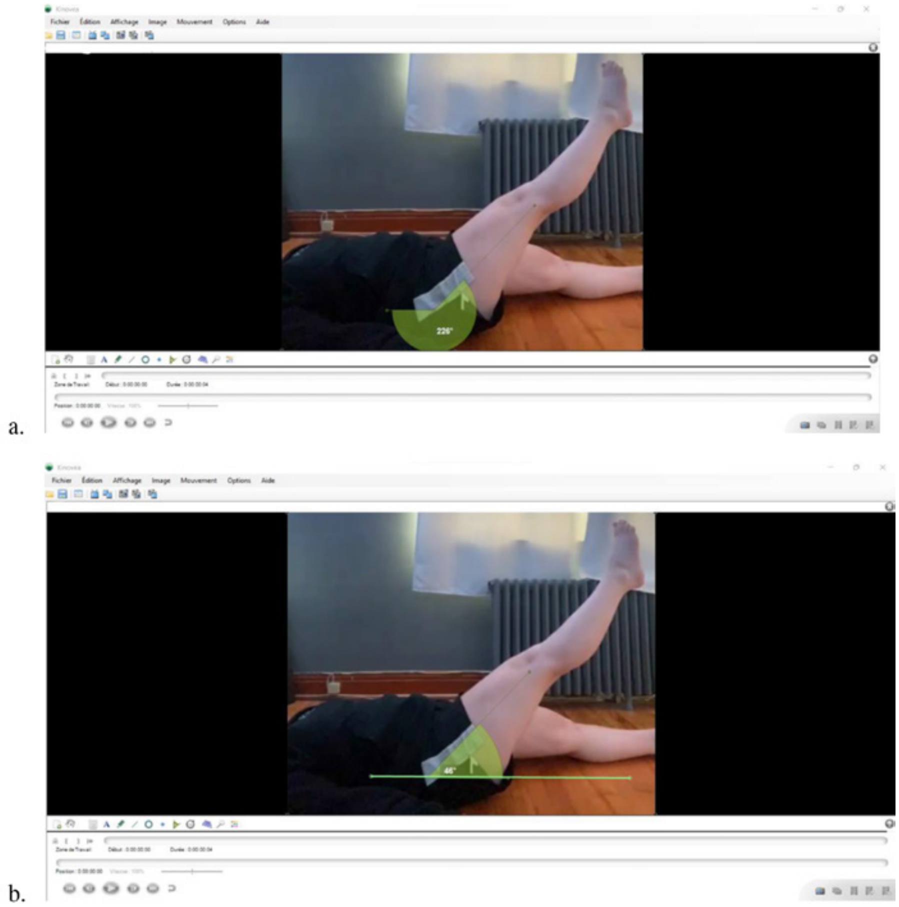

Figure 3. View of the hip flexion measured with the virtual goniometer. a) If the static branch of the goniometer is directly placed on bony landmark the reference of 180° needs to be subtracted. b) To obtain direct measurement, a line can be prolonged from the proximal segment and the goniometer positioned on this line.

Statistical analysis

Statistical analyses were performed using IBM SPSS Statistics version 26 for Windows (IBM Corp., Armonk, NY, USA). Descriptive statistics were performed using non-parametric statistic when the data were not normally distributed and parametric statistics when they were normally distributed. The inter-rater reproducibility was assessed using intraclass correlation coefficient (ICC) and associated 95% CI for each joint and overall. The inter-rater reproducibility was calculated based on single measurement, absolute-agreement, 2-way random-effects model ICC[20-22]. The two-way random-effect model was selected as this model is appropriate for evaluating rater-based clinical assessment methods. The two-way random effects model was used as the intent was to generalize the reliability results to any raters as opposed to the two-way mixed effects model, which only represents the specific raters involved in the reliability experiment. Absolute agreement definition was selected as the goal was to verify if different raters assign the same score to the same subject. The intra-rater repeatability (measure 1 vs. measure 2) was assessed for both raters using ICC with a two-way mixed effect model, mean of measurements type and absolute agreement definition[20]. The two-way mixed effect model was selected as this model is appropriate for testing multiple scores from the same rater. Absolute agreement definition was used for the same reason as for inter-rater ICC. A mean of measurements type was selected for the intra-rater ICC as the analysis was based on multiple measurements. Coefficients of variation (CV) were calculated based on formula 1 to complement the ICC as CV is often used to assess variability[23]. Paired t-tests were used to assess systematic bias by calculating the change in the mean between the two measures of each rater.

Formula 1. Coefficient of variation (in %):

$$\text{Coefficient of variation} = \left(\frac{\text{Standard deviation of the differences between measures 1 and 2}}{\text{mean of all absolute measurements}}\right) * 100$$

Table 1. Participants' demographic information.

Participant ID	Age	Sex	Location of joint contractures [a]	Mobility status
1	16	F	Shoulder, elbow, wrist, hand, knee, ankle, foot, spine	Non-ambulatory
2	21	M	Shoulder, elbow, wrist, hand, hip, knee, ankle, foot	Household
3	12	M	Shoulder, elbow, wrist, knee, ankle, foot	Community
4	11	M	Elbow, wrist, hand, hip, knee, foot	Household (limited)
5	17	F	Elbow, wrist, hand, ankle, foot	Community
6	20	M	Shoulder (left), hip (right), knee, ankle	Community
7	17	M	Shoulder, elbow, wrist, knee, ankle, foot	Community (limited)
8	15	F	Hip, knee, ankle	Community
9	16	M	Shoulder, elbow, wrist, hand, hip, knee, ankle	Community

[a] All contractures but otherwise specified were bilateral.

Table 2. Median and 95% confidence interval (CI) for upper limb range of motion measured by each rater with the average of absolute difference of measurement.

Movement (number[a])		1st rater			2nd rater			Significance (1/2/both)*
		1st time Median (95% CI) in degree°	2nd time Median (95% CI) in degree°	Average of absolute difference of measures (in %)	1st time Median (95% CI) in degree°	2nd time Median (95% CI) in degree°	Average of absolute difference of measures (in %)	
Shoulder	Abduction (n=26)	155.5 (141;163)	154.0 (137; 159)	3%	148.0 (120; 165)	158.0 (146; 167)	6%	a/b/a
	Flexion (n=16)	105.5 (41; 151)	106.5 (35; 145)	4%	109.0 (38; 149)	109.5 (38; 143)	2%	a/a/a
	Extension (n=13)	50.0 (18, 66)	49.0 (18, 65)	6%	49.0 (21; 70)	43.0 (19; 71)	4%	a/a/a
Elbow	Flexion (n=27)	143.0 (96; 155)	144.0 (97; 155)	3%	141.0 (97; 151)	141.0 (100; 154)	4%	a/a/a
	Extension (n=27)	-4.0 (-11; 0)	-5.0 (-12; -2)	10%	-3.0 (-12; 0)	-3.0 (-10; 0)	10%	a/a/a
Forearm	Pronation (n=22)	84.5 (76; 98)	85.0 (78; 93)	7%	87.5 (83; 98)	89.5 (79; 98)	2%	a/a/a
	Supination (n=22)	68.5 (56; 84)	72.0 (56; 84)	6%	74.5 (63; 92)	76.5 (68; 92)	6%	a/a/b
Wrist	Flexion (n=28)	78.5 (67; 85)	77.0 (67; 85)	5%	73.5 (61; 90)	77.0 (66; 86)	8%	a/a/a
	Extension (n=28)	25.0 (-7; 50)	22.5 (-6; 53)	8%	14.0 (-9; 52)	21.5 (-4; 47)	10%	a/a/a

**Significance level of the difference between ROM measure 1 vs. 2 for rater 1 and 2 (intra-rater) and for average measures between raters (inter-rater). [a] Significance> 0.05; [b] Significance <0.05.*

Results

Participants

Three female and six male participants with a median age of 15.9 years (range: 11.3 to 20.8 years) were included in the study (Table 1). Seven participants with pre-and post-intervention ROM data and two participants who only had pre-intervention ROM data were analysed. The ROM are presented in Tables 2 and 3 as median and 95% confidence interval (CI) for consistency. The number of joints with ROM measurements are reported in all Tables. On a possibility of 544 ROM measurements ((17 ROM measurements*7participants*2 sides*2 times)+(17 ROM measurements*2 participants*2 sides*1 time)), 91 (16.7%) were not assessed, 13 (2.39%) were assessed in the wrong plane of movement and 8 (1.47%) were not assessed using the same method. Among those not assessed, 40 ROM measurements were not assessed because the participant

Table 3. Median and 95% confidence interval (CI) for lower limb range of motion measured by each rater with the average of absolute difference of measurement.

Joints	Movement (Number[a])	1st rater			2nd rater			Significance (1/2/both)*
		1st time Median (95% CI) in degree°	2nd time Median (95% CI) in degree°	Average of absolute difference of measures (in %)	1st time Median (95% CI) in degree°	2nd time Median (95% CI) in degree°	Average of absolute difference of measures (in %)	
Hip	Flexion (n=30)	106.0 (100; 113)	103.0 (94; 112)	5%	104.5 (95; 114)	104.5 (93; 114)	3%	b/a/a
	Extension (n=23)	16.0 (10; 22)	15.0 (8; 17)	19%	12.0 (-14; 16)	13.0 (-10; 18)	18%	a/a/a
	Internal rotation (n=26)	20.0 (10; 29)	21.5 (11; 28)	9%	24.0 (10; 30)	20.5 (10; 30)	6%	a/a/a
	External rotation (n=26)	30.0 (21; 49)	30.0 (18; 49)	6%	34.0 (19; 50)	35.0 (18; 49)	4%	a/a/a
Knee	Flexion (n=32)	96.5 (66; 120)	100.5 (65; 122)	3%	92.0 (62; 120)	103.0 (59; 123)	4%	b/b/a
	Extension (n=32)	-20.0 (-37; -8)	-21.0 (-35; -9)	8%	-18.5 (-29; -9)	-18.0 (-32; -7)	9%	a/a/c
Ankle	Dorsiflexion (n=28)	-20.0 (-28; -5)	-13.5 (-24; -4)	20%	-21.0 (-27; -7)	-21.5 (-29; -7)	10%	d/a/d
	Plantarflexion (n=28)	31.5 (25; 40)	27.5 (20; 36)	15%	21.0 (13; 37)	37.0 (23; 46)	26%	d/c/a

**Significance level of the difference between ROM measure 1 vs. 2 for rater 1 and 2 (intra-rater) and for average measures between raters (inter-rater). [a] Significance> 0.05; [b] Significance <0.05; [c] Significance <0.01; [d] Significance ≤ 0.001.*

Table 4. Intra-rater repeatability and coefficient of variation (%) for upper limb range of motion.

Joints	Movement (Number[a])	1st rater			2nd rater		
		ICC	95% CI	CV	ICC	95% CI	CV
Shoulder	Abduction (n=26)	0.996	0.992; 0.998	4%	0.976	0.935; 0.990	9%
	Flexion (n=16)	0.994	0.983; 0.998	5%	0.998	0.994; 0.999	3%
	Extension (n=13)	0.972	0.913; 0.991	12%	0.994	0.979; 0.998	6%
Elbow	Flexion (n=27)	0.994	0.986; 0.997	4%	0.985	0.968; 0.993	6%
	Extension (n=27)	0.995	0.990; 0.998	14%	0.997	0.995; 0.999	13%
Forearm	Pronation (n=22)	0.861	0.698; 0.939	10%	0.988	0.972; 0.995	3%
	Supination (n=22)	0.932	0.846; 0.971	10%	0.925	0.830; 0.968	11%
Wrist	Flexion (n=28)	0.985	0.969; 0.993	6%	0.957	0.909; 0.980	12%
	Extension (n=28)	0.997	0.992; 0.998	10%	0.990	0.978; 0.995	17%

*[a] Number of data included for analysis with a possible maximum of 32: (7 participants * 2 sides * 2 times) + (2 participants * 2 sides * 1 time).*

had no upper limb active ROM. Among those excluded because they were in the wrong plane of movements, 12 were for shoulder flexion.

Intra-rater repeatability

The intra-rater repeatability varied from poor to excellent among the different joint movements and can be found in Tables 4 and 5 for both raters. The overall intra-rater ICC was 0.997 (95% CI:0.996 to 0.997) for the first rater and 0.993 (95% CI:0.992 to 0.994) for the second rater. For the first rater, the lowest ICC was for the forearm pronation with an ICC of 0.861 (95% CI: 0.698 to 0.939) and the highest ICC was for wrist extension with an ICC of 0.997 (95% CI: 0.992 to 0.998). For the second rater, the lowest ICC was for

Table 5. Intra-rater repeatability and coefficient of variation (%) for lower limb range of motion.

Joints	Movements (Number[a])	1st rater			2nd rater		
		ICC	95% CI	CV	ICC	95% CI	CV
Hip	Flexion (n=30)	0.957	0.895; 0.981	6%	0.955	0.909; 0.978	6%
	Extension (n=23)	0.974	0.937; 0.989	24%	0.970	0.932; 0.987	27%
	Internal rotation (n=26)	0.991	0.980; 0.996	12%	0.995	0.988; 0.998	8%
	External rotation (n=26)	0.991	0.980; 0.996	8%	0.993	0.985; 0.997	7%
Knee	Flexion (n=32)	0.996	0.991; 0.998	3%	0.984	0.965; 0.992	8%
	Extension (n=32)	0.994	0.989; 0.997	10%	0.991	0.981; 0.995	13%
Ankle	Dorsiflexion (n=28)	0.969	0.64; 0.991	15%	0.985	0.967; 0.993	16%
	Plantarflexion (n=28)	0.929	0.606; 0.977	13%	0.740	0.410; 0.884	36%

*[a] Number of data included for analysis with a possible maximum of 32: (7 participants * 2 sides * 2 times) + (2 participants * 2 sides * 1 time).*

Table 6. Inter-rater reproducibility for upper limb range of motion measurement.

Joints	Movement (Number[a])	ICC	95% CI	Significance
Shoulder	Abduction (n=26)	0.997	0.994; 0.999	<0.001
	Flexion (n=16)	0.998	0.994; 0.999	<0.001
	Extension (n=13)	0.996	0.986; 0.999	<0.001
Elbow	Flexion (n=27)	0.998	0.996; 0.999	<0.001
	Extension (n=27)	0.988	0.974; 0.995	<0.001
Forearm	Pronation (n=22)	0.410	-0.392; 0.753	0.115
	Supination (n=22)	0.832	0.565; 0.932	<0.001
Wrist	Flexion (n=28)	0.976	0.947; 0.989	<0.001
	Extension (n=28)	0.994	0.986; 0.997	<0.001

*[a] Number of data included for analysis with a possible maximum of 32: (7 participants * 2 sides * 2 times) + (2 participants * 2 sides * 1 time).*

Table 7. Inter-rater reproducibility for lower limb range of motion measurement.

Joints	Movement (Number[a])	ICC	95% CI	Significance
Hip	Flexion (n=30)	0.982	0.962; 0.991	<0.001
	Extension (n=23)	0.921	0.816; 0.966	<0.001
	Internal rotation (n=26)	0.987	0.971; 0.994	<0.001
	External rotation (n=26)	0.993	0.983; 0.997	<0.001
Knee	Flexion (n=32)	0.996	0.993; 0.998	<0.001
	Extension (n=32)	0.995	0.984; 0.998	<0.001
Ankle	Dorsiflexion (n=28)	0.979	0.922; 0.992	<0.001
	Plantarflexion (n=28)	0.954	0.900; 0.979	<0.001

*[a] Number of data included for analysis with a possible maximum of 32: (7 participants * 2 sides * 2 times) + (2 participants * 2 sides * 1 time).*

ankle plantarflexion with an ICC of 0.740 (95% CI: 0.410 to 0.884) and the highest was for shoulder flexion with an ICC of 0.998 (95% CI: 0.994 to 0.999). The CV ranged between 3% and 24% with an overall CV of 9% for the first rater, with the knee flexion being the lowest and the hip extension the highest. For the second rater, the CV ranged between 3% and 36% with an overall CV of 11%. The shoulder flexion had the lowest CV and the ankle plantarflexion the highest.

Inter-rater reproducibility

The inter-rater reproducibility varied from poor to excellent among joint movements and are shown in Tables 6 and 7. The highest ICC was found for the shoulder flexion and elbow flexion with an ICC of 0.998 (95% CI: 0.994 to 0.999) and 0.998 (95% CI: 0.996 to 0.999) respectively. Two ICCs were below 0.900: the forearm pronation had an ICC of 0.410 (95% CI: -0.392 to 0.753) and the forearm supination had an ICC of 0.832 (95% CI: 0.565 to 0.932). The overall ICC was 0.995 (95% CI: 0.994; 0.996, $p<0.001$).

Discussion

In the present study, the measurement repeatability and reproducibility of using a virtual goniometer to measure ROM of the upper and lower limbs of youths with AMC based on screen captures was assessed. This study suggests that despite challenges associated with the patient condition (e.g. skin webbing, limited ROM) and the uncontrolled environment, virtual goniometry is a repeatable and reproducible clinical assessment tool for measuring ROM across most joints. All joints with long lever-arms had good to excellent reliability (e.g. shoulder extension, knee flexion, hip internal rotation). On the other hand, the lowest repeatability and reproducibility values were reported for joints with short lever-arms such as forearm pronation and supination, and ankle dorsiflexion and plantarflexion. Therefore, further investigation with improved techniques or more practice is needed to confirm ROM measurement reliability across joints with short lever-arms before moving forward with studies of procedural reliability and validity.

A thorough search of the relevant literature yielded only two related articles that assessed measurement repeatability and reproducibility of virtual goniometry. Both studies assessed healthy adults simulating an impairment in a controlled (clinical) context[13,14]. Their results showed moderate to excellent repeatability and reproducibility for the wrist flexion (intra-rater $ICC_{2,1}$: 0.70-0.97; inter-rater $ICC_{2,2}$: 0.83) and extension (intra-rater $ICC_{2,1}$: 0.65-0.84; inter-rater $ICC_{2,2}$: 0.69), and knee flexion (intra-rater $ICC_{2,1}$: 0.91-1.00; inter-rater $ICC_{2,2}$: 0.90-1.00) and extension (intra-rater $ICC_{2,1}$: 0.66-1.00; inter-rater $ICC_{2,2}$: 0.64-0.96). These results are similar to what has been reported in the current study despite the challenges associated with the patient condition's (e.g. skin webbing, body compensation during ROM assessment). Thus, the current study suggests that added challenges associated with the youth's condition seem to have had minimal impact on joint ROM measurements when repeated on captured images.

Amongst all joints tested there were two specificities that stood out and led to increased variability in the measurement. First, joints with short lever-arms (forearm pronation and supination, ankle dorsiflexion and plantarflexion), showed generally lower ICCs than those with longer lever-arms. Amongst those short lever-arm joints, only wrist flexion and extension had excellent repeatability and reproducibility. This is similar to the study by Mehta et al. who reported moderate to excellent reliability for wrist flexion (intra-rater $ICC_{2,1}$: 0.70-0.97; inter-rater $ICC_{2,2}$: 0.83) and extension (intra-rater $ICC_{2,1}$: 0.65-0.0.84; inter-rater $ICC_{2,2}$: 0.69). All the other short lever-arm joints of the current study showed poor to moderate intra or inter-rater ICCs. There are two potential explanations as of why short lever-arms led to lower reliability than those with longer lever-arms. First, the ROM of these joints are generally smaller which affects the calculation of the ICC. This is due to the fact that ICCs reflect whether a subject maintains their rank within the study population between sessions and thus is influenced by the spread of values found within a study population[24]. Therefore, if all values are within a span of 10° it is harder to keep the rank than if the span is larger. Second, those short lever-arms often do not provide clear visible anatomical landmarks compared to longer lever-arms (e.g. ankle dorsiflexion vs elbow flexion), which may lead to an increased difficulty in measuring the ROM.

Second, coefficients of variation were generally larger when measuring joints in extension compared to those in flexion. It was subjectively harder to measure movements in extension rather than in flexion, more so when the participants presented with skin webbing (Figure 1), as the axes of joint rotation were less visible. Thus, this could be contributing to larger CV. However, this is unlikely given that this subjective difficulty did not lead to lower ICCs for joints showing larger CV. Another possible explanation for these larger CVs is that they are strongly influenced by the average of the ROM capture as the variability is divided by the mean of the measurements. Therefore, for example, if the average ROM for hip extension is 5°, then the CV will almost certainly be larger than the ROM for hip flexion of 103° if the standard deviation is similar, which is the case for hip flexion and extension with a standard deviation of about 22° for hip flexion and extension (Table 3). However, only hip extension (average: 4.64°; SD: 23.0°) and ankle dorsiflexion (average: -16.8°, SD: 19.9°) were above 15% for both raters, which is the most common cut-off found in the literature[25].

Most, if not all, previous studies showing that virtual goniometry is repeatable and reproducible were conducted in controlled environments, but this current study was conducted in the participants' home or school[13,14]. Therefore, inherent challenges including limited space, bad lighting (too little or too much), and difficulty with camera alignment were encountered. Despite these added challenges, measurement repeatability and reproducibility have been found to be similar to what was found in previous studies using controlled environment[13,14]. This suggests that the environment does not have a strong impact on measurement reliability if some guidelines are followed (e.g. portable devices, contrasting clothes).

In the above-mentioned studies, the ROM measurement were only assessed in the sagittal plane i.e., flexion and extension. In the current study, repeatability and reproducibility were found to have good to excellent agreement in other planes of motion. Thus, this study shows that in addition to sagittal plane motion, frontal and transverse planes motion also provide reliable data when the participants are positioned in the proper plane of movement.

In the current study, the procedural reliability and the validity of the virtual goniometer were not evaluated. It is expected that measurement reliability will be higher than procedural reliability, as differences in patient positioning between testing sessions and fluctuations in performance are eliminated as potential sources of error. When compared to a study from Johansen et al. that assessed within-day procedural repeatability among children with cerebral palsy, reliability results were similarly moderate-to-excellent (intra-rater $ICC_{2,1}$: 0.663; inter-rater $ICC_{2,1}$: 0.626) for ankle dorsiflexion[26]. However, as expected, the reproducibility results from the current study were excellent, and higher than the moderate reproducibility ($ICC_{2,1}$: 0.626) found in Johansen et al. on individuals with cerebral palsy for ankle dorsiflexion[26]. Another study also assessed within-day procedural reproducibility using photograph-based goniometry among participants seen in an orthopedic hand clinic and obtained similar results compared to the current study with good-to-excellent reproducibility for joints with longer lever-arms ($ICC_{N/A}$: 0.83 (elbow flexion) – 0.96 (elbow extension)) and lower reliability for shorter lever-arms ($ICC_{N/A}$: 0.28 (radial deviation and forearm pronation) – 0.94 (forearm supination))[27]. This suggests that procedural variability does not seem to have a strong effect on reliability of virtual ROM measurement. The tendency of lower ICC for joints with shorter lever-arms seems to also apply for the validity. Three studies assessed within-day validity comparing in-person measurement using a universal goniometer with virtual goniometer with people with Parkinson disorder[28], children with cerebral palsy[26] and healthy participants[13]. Similar to what was found in the current study, authors found moderate to excellent validity for joint with longer lever-arms ($ICC_{2,1}$: 0.49 (elbow flexion)-1.00 (knee flexion and extension)) and lower validity for joints with shorter lever-arms ($ICC_{2,1}$: 0.121 (ankle dorsiflexion)-0.86 (wrist flexion)).

Strengths and Limitations

The sample size is small, but given that both sides were assessed at two independent time points, the sample ranged from 13 to 32 measurements for each ROM measurement. Based on Glüer, Blake et al. (1995), a minimum of 27 degrees of freedom is recommended to determine reproducibility[29]. This recommendation has been met for 9 out of the 17 movements assessed during the study. The movements that did not meet this recommendation were the following (degree of freedom): shoulder abduction (26), shoulder flexion (16), shoulder extension (13), forearm pronation (22), forearm supination (22), hip extension (23), hip internal rotation (26) and hip external rotation (26). However, based on the Q-Q plots, 5 out of the 8 measurements that did not have sufficient degrees of freedom seemed to be relatively normally distributed (shoulder flexion, shoulder extension, forearm pronation, forearm supination and hip external rotation). The assessors involved in the remote assessment of the ROM did not have a standardized training on how to take proper measurements remotely that may have affected the testing position, e.g. shoulder flexion performed in frontal view instead than in sagittal view, and consequently some data was lost due to not meeting the proper requirements for measurement. The raters involved in the ROM measurements did not have prior experience with the use of virtual goniometer for ROM measurements before the start of the study, which may have led to certain discrepancies in their measures. However, a training with three patients was provided to both raters to compensate for the lack of experience with virtual goniometry. In addition, this may better represent the reality of a clinician who will start using a virtual goniometer in his or her clinical practice. Moreover, despite minimal experience and training, the measurements were highly reliable suggesting that it did not greatly affect the results of the study. Raters also used existing screen captures from the telerehabilitation study, which created a potential source of bias in favor of positive results as they used the same screen capture. This study evaluated the repeatability and reproducibility of the measurement with a virtual goniometer. Future studies should assess the procedural reliability by evaluating the whole process including participants positioning. A limitation in the current study involves validity of virtual goniometer compared to in-person measurements. Although the validity has been already evaluated in previous studies, it has not been explored for all ROM, with complex clinical populations and compared with target environments. In addition, future studies should assess the effect of camera angle and distance from the camera on the perceived range of motion. Comparison with in-person measurement was not possible due to the main study having been conducted completely remotely. Finally, active ROM was taken in the context of this study as it was more meaningful to create a home-based exercise program, but future studies should assess the possibility of assessing passive ROM with the help of a caregiver.

In the current study, the most common cut-offs for ICC found in the literature were used and were as follows; excellent: ≥0.90, good: 0.75-0.90, moderate: 0.50-0.75 or poor: <0.50[20]. However, no hard cut-offs were found in the literature, and the acceptable level should depend on the context of the use and the degree of acceptable precision in the measurement. The acceptable level may vary depending on if the virtual goniometer is being used to assess for surgical indications, for follow-ups and re-evaluation of ROM improvement following a conservative intervention, or as a research tool.

This study is one of the first to analyze the repeatability and reproducibility of the use of a virtual goniometer in a natural setting with a complex clinical population. Participants with physiological limitations to their ROM, skin webbing and various contractures were included in the study, which led to a better representativeness of the results. Participants were assessed while they were at home in a real remote setting, which better corresponds to the telerehabilitation context and increases the applicability of the results. The data may further contribute to and facilitate the clinical implementation of virtual goniometry with other complex clinical populations in the future.

Recommendations

Based on what was experienced in the study, the following recommendations apply:

- To decrease the possibility of compensation due to lack of active ROM (e.g. lateral bending of the trunk during shoulder abduction), take bilateral ROM simultaneously when possible, considering that bilateral ROM measurement is feasible with the screen capture feature.
- To increase the chance of obtaining good images of the different ROM, it is recommended to ask the participant to wear contrasting clothes, to have enough space around them to perform the movement in the camera frame, to ensure proper lighting and to use a portable device (e.g., laptop, tablet) to allow for flexible camera placement.
- To avoid loss of data, training clinical staff on how to position the patient in front of the camera and to use a data collection sheet indicating the plane of movement (Supplementary Table) is recommended because remote ROM measurement is not as intuitive as in person.
- To improve the measurement reliability of movements with shorter lever-arms, such as forearm pronation and ankle plantarflexion, it could help to take the measurement closer to the camera and make sure the screen capture is as clear as possible.

Conclusions

In summary, this study demonstrated that remote measurement of active ROM with a virtual goniometer is a promising approach. Besides the following exceptions (i.e., forearm pronation and supination, and ankle dorsiflexion and plantarflexion; mostly short lever-arms joints), the current study suggests that this measurement approach of evaluating screen captures using virtual goniometry seems reproducible and repeatable in youth with AMC. Despite challenges that arose with the target population and remote assessment in non-clinical environments (e.g. lighting, space and positioning) repeatability and reproducibility was possible. However, further steps are needed before clinically implementing the virtual goniometer. Future studies should aim to enhance measurement reliability of movements with shorter lever-arms (i.e. forearm supination and pronation, ankle dorsiflexion and plantarflexion), establish procedural reliability, and determine validity of the proposed method for youth with complex neuro-musculoskeletal conditions.

Ethics approval

The project received ethical approval from the McGill University Faculty of Medicine Institutional Review Board (#A08-B38-18B) in October 2018.

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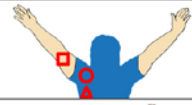
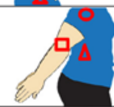
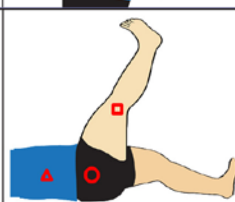
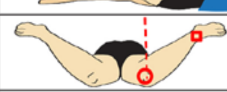
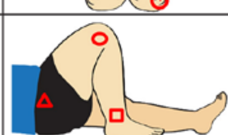
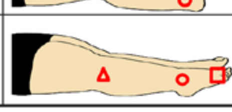
Supplementary File.

Joint	Movement	Plane of movement	Visual	Right (in °)	Left (in °)	Tips
Shoulder	Abduction	Frontal (both side simultaneously)				
	Flexion	Sagittal				
	Extension	Sagittal				
Elbow	Flexion	Sagittal				
	Extension	Sagittal				
Forearm	Pronation	Frontal (both side simultaneously)				Ask the person to hold a pen while doing the movement. It is easier to measure with the virtual goniometer.
	Supination	Frontal (both side simultaneously)				
Wrist	Flexion	Sagittal				If passive wrist ROM is needed, ask the person to put the palm of their hands together (extension) or the reverse part of their hand (flexion) and go as down as possible until it starts not touching the other.
	Extension	Sagittal				
Hip	Flexion	Sagittal				Ask the person to lay down on the floor with the camera at the same level
	Extension	Sagittal				
	Internal rotation	Transverse (both side simultaneously)				
	External rotation	Transverse (both side simultaneously)				
Knee	Flexion	Sagittal				
	Extension	Sagittal				
Ankle	Dorsiflexion	Sagittal				
	Plantarflexion	Sagittal				

○ Fulcrum
△ Bony landmark for the stationary arm
□ Bony landmark for the moving arm

Upper extremity range of motion

Shoulder abduction
The individual positions themself in front of the camera. They will abduct the shoulder by moving their arm laterally away from the trunk.
Fulcrum: Anterior aspect of the acromial process
Proximal arm: Parallel to the midline of the sternum
Distal arm: Midline of the humerus[1]

Shoulder flexion
The individual positions themself sideways to the camera. To limit the possibility of compensation due to remote assessment, they will flex both shoulders simultaneously by bringing their hands as high as possible forward. Their hands need to be in a neutral position. The measurement is done on the side that is the nearest of the camera.
Fulcrum: Lateral aspect of the greater tubercle
Proximal arm: Parallel to the midaxillary line of the thorax
Distal arm: Lateral midline of the humerus

Shoulder extension
The individual positions themself sideways to the camera. To limit the possibility of compensation due to remote assessment, they will extend both shoulders simultaneously by bringing their hands as high as possible backward. Their hands need to be in a neutral position. The measurement is done on the side that is the nearest of the camera.
Fulcrum: Lateral aspect of the greater tubercle
Proximal arm: Parallel to the midaxillary line of the thorax
Distal arm: Lateral midline of the humerus

Elbow flexion
The individual positions themself sideways to the camera with the forearm in full supination. To limit the possibility of compensation due to remote assessment, they will bend both elbows by moving the hands toward their shoulders. The measurement is done on the side that is the nearest of the camera.
Fulcrum: Lateral epicondyle of the humerus
Proximal arm: Lateral midline of the humerus
Distal arm: Lateral midline of the radius

Elbow extension
The individual positions themself sideways to the camera with the forearm in full supination. To limit the possibility of compensation due to remote assessment, they will extend both elbows by moving both hands away from the shoulder. The measurement is done on the side that is the nearest of the camera.
Fulcrum: Lateral epicondyle of the humerus
Proximal arm: Lateral midline of the humerus
Distal arm: Lateral midline of the radius

Forearm pronation
The individual positions themself in front of the camera with the elbow at 90°. They will turn their palm down as much as possible. To limit the possibility of compensation due to remote assessment, they will perform the movement bilaterally at the same time.
Fulcrum: Laterally and proximally to the ulnar styloid process
Proximal arm: Parallel to the anterior midline of the humerus
Distal arm: Dorsal aspect of the forearm, proximal to the styloid processes of the radius and ulna

Forearm supination
The individual positions themself in front of the camera with their elbow at 90°. They will turn their palm up as much as possible. To limit the possibility of compensation due to remote assessment, they will perform the movement bilaterally at the same time.
Fulcrum: Medially and proximally to the ulnar styloid process
Proximal arm: Parallel to the anterior midline of the humerus
Distal arm: Ventral aspect of the forearm, proximal to the styloid processes of the radius and ulna

Wrist flexion
The individual positions themself sideways to the camera with their elbow at 90° and the palm of their hand facing the floor. They will flex their wrist as much as possible.
Fulcrum: Lateral aspect of the wrist
Proximal arm: Lateral midline of the ulna
Distal arm: Lateral midline of the fifth metacarpal

Wrist extension
The individual positions themself sideways to the camera with their elbow at 90° and the palm of their hand facing the floor. They will extend their wrist as much as possible.
Fulcrum: Lateral aspect of the wrist
Proximal arm: Lateral midline of the ulna
Distal arm: Lateral midline of the fifth metacarpal

Lower extremity range of motion

Hip flexion
The individual positions themself lying supine on the floor with the knees extended sideways to the camera. They will flex their hip by lifting the leg off the floor as far as possible.
Fulcrum: Lateral aspect of the hip joint
Proximal arm: Lateral midline of the pelvis
Distal arm: Lateral midline of the femur

Hip extension
The individual positions themself lying prone on the floor with the knees extended sideways to the camera. They will extend their hip by lifting the leg off the floor as far as possible, without excessively arching their back.
Fulcrum: Lateral aspect of the hip joint
Proximal arm: Lateral midline of the pelvis
Distal arm: Lateral midline of the femur

Hip internal rotation
The individual positions themself lying prone on the floor, facing back to the camera and with the knees flexed at 90°. They will rotate their hips internally by bringing their feet as externally as possible. As it is not possible to stabilize the pelvis during a remote assessment, it is recommended to perform this test bilaterally at the same time.
Fulcrum: Anterior aspect of the patella
Proximal arm: Perpendicular to the floor
Distal arm: Anterior midline of the lower leg

Hip external rotation
The individual positions themself lying prone on the floor, facing back to the camera and with the knees flexed at 90°. They will rotate their hips externally by bringing their feet as internally as possible. As it is not possible to stabilize the pelvis during a remote assessment, it is recommended to perform this test bilaterally at the same time. The leg that is the nearest to the camera is the leg that will be measured and the test will be repeated again with the other leg in front.
Fulcrum: Anterior aspect of the patella

[1] All explanations from this Supplementary File are based on Norkin CC, White DJ. Measurement of joint motion: a guide to goniometry. 4th ed. ed. Philadelphia: F.A. Davis; 2009

Proximal arm: Perpendicular to the floor
Distal arm: Anterior midline of the lower leg

Knee flexion

The individual positions themself supine on the floor with the knees extended sideways to the camera. They will bring their foot as close as possible to their thigh.
Fulcrum: Lateral epicondyle of the femur
Proximal arm: Lateral midline of the femur
Distal arm: Lateral midline of the fibula

Knee extension

The individual positions themself supine on the floor sideways to the camera. They will place their foot on a roll of towels or a pillow to allow for full knee extension.
Fulcrum: Lateral epicondyle of the femur
Proximal arm: Lateral midline of the femur
Distal arm: Lateral midline of the fibula

Ankle dorsiflexion

The individual positions themself supine on the floor sideways to the camera with their knee fully extended. They will lift their toes toward their head.
Fulcrum: Lateral aspect of the lateral malleolus
Proximal arm: Lateral midline of the fibula
Distal arm: Parallel to the lateral aspect of the fifth metatarsal

Ankle plantarflexion

The individual positions themself supine on the floor sideways to the camera with their knee extended. They will point their toes toward the floor.
Fulcrum: Lateral aspect of the lateral malleolus
Proximal arm: Lateral midline of the fibula
Distal arm: Parallel to the lateral aspect of the fifth metatarsal

Additional considerations

- Ask to align the camera as much as possible (e.g. if possible position the camera on the floor to be align with the hip flexion/ extension movement
- A movement should be excluded for the following reasons:
  Extreme physiological compensations accompanying the desired joint movements that render too difficult to measure in the right plane of movement
  Improper plane of movement (e.g. shoulder flexion perform in frontal plane)
- In order to obtain a direct measure without the need of performing calculation, a line can be prolonged from the proximal segment to position the virtual goniometer (refer to figure 3b in the manuscript).