Research Article

A PILOT STUDY INVESTIGATING THE RELIABILITY AND FEASIBILITY OF MEASURING TIBIAL LENGTH AND RECUMBENT LENGTH IN CHILDREN AND YOUNG ADULTS WITH CEREBRAL PALSY?

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ABSTRACT

Aim:
To investigate the validity and feasibility of using tibial and recumbent length measurements in children with Cerebral Palsy (CP) in the outpatient clinic setting.

Method:
Participants with a primary diagnosis of Cerebral Palsy (CP) aged between 2-20 years of age (n=38) were recruited from a Paediatric Orthopaedic Rehabilitation outpatient clinic or a school for children with CP.

Intraobserver and interobserver reliability for tibial and recumbent length were measured using standardised methods. Agreement between recumbent length and standing height was assessed.

Results: For tibial length the Technical Error (TE) for intraobserver was 0.6cm and the Coefficient of Variation (CV) was 2.1% (n=31); the interobserver TE was 0.9cm and CV 3.1% (n=33).

For recumbent length the intraobserver TE was 2.0cm and the CV was 1.4% (n=26); the interobserver TE was 2.3cm and CV 1.6% (n=30).

For fourteen subjects who were able to stand, the mean (SD) difference between measured and recumbent length was 3.7 (3.2) cm ($p<0.001$).

No children declined any of the tibial measurements. Two children declined their first recumbent measurement and three declined a repeated measurement.

Conclusion:
Tibial length is a useful measure of growth in children with CP who are unable to stand for height. It is feasible to measure in the outpatient clinic (OPC) setting and acceptable by children. Further standardisation of methods and training however is required to improve measurement reliability and this should be retested.

Keywords: cerebral palsy; height; length; growth

INTRODUCTION:

Cerebral palsy (CP) is one of the most common causes of disability with a prevalence of 2 to 3 per thousand life births every year[1].

It is difficult to monitor the growth of children with CP because measurement of height/recumbent length is often inaccurate and unreliable due to contractures and scoliosis. Also standard growth charts may not be appropriate to use given the differences in growth patterns of children particularly in children severely affected[2].

It is however important to monitor the growth of children with CP because:

- It has been reported that growth patterns in children with CP are associated with overall health and social participation[3];
- Feeding difficulties can be partly responsible for the deficits in growth and these can be modified;
- Endocrine and metabolic abnormalities can present with poor growth;
- Height/length are useful to calculate body mass index and predict energy requirements, and are also useful to predict reference ranges for pulmonary function and glomerular filtration rate.

In New Zealand there is no standardised approach to measuring children with moderate to severe cerebral palsy[4]. Segmental measures such as knee height, tibial and ulna length are frequently used as a proxy for standing height in children with severe cerebral palsy. They have been shown to be reproducible and to correlate well with height in both typically developing children and children with CP[5,6,7]. Tibial length can be plotted directly onto growth charts created from data collected as part of The Northern American Growth in Cerebral Palsy Project (NAGCPP)[3,8]. Tibial length can also be used in prediction equations to estimate standing height although there are large limits of agreement with standing height[5,9,10,11]. Tibial length has been shown to be reproducible and also have a correlate well with height in both typically developing children and children with CP [(5,6).]

Recently Haapala et al used continuous segmental measures to measure recumbent length in 137 children and young adults (aged 2-25 years) with CP in the United States and found good inter- and intraobserver reliability[12]. Interobserver reliability however was only carried out on a small number of children (n=16) with Gross Motor Function Classification System (GMFCS) level unreported.

In this study we wanted to investigate the reliability and feasibility of a technique that would allow measurement of linear growth over time, one that does not involve special equipment or highly

technical training and that is suitable for use in an outpatient setting. Also we wanted the measuring process to be acceptable to the child.

We therefore chose to measure tibial length, which requires no special equipment and has previously been shown to have good reliability[7], as well as recumbent length using the methods outlined in Haapala et al [12].

Methods

Participants:

This was a cross sectional study of a convenience sample of children with a primary diagnosis of CP selected from a Paediatric Orthopaedic Rehabilitation OPC and a Special School for children with CP in Auckland, New Zealand during 2016. Children were eligible to participate if they had a primary diagnosis of CP and were aged 2-20 years. They were excluded if they had a history of a genetic, metabolic or neurodegenerative disorder.

For children who attended the Special School written consent was obtained from caregivers prior to the day of measuring. Participant information with a consent form was sent home and the caregivers were asked to return it to the school or to the investigators. For the children who attended the outpatient clinic written consent was obtained at the time of their appointment. They were approached by the primary investigator and given participant information.

Where possible assent or consent was also obtained from participants. All children were seen in a private room and were able to withdraw from the study at any stage and opt out of any measurements when they indicated to parent or teacher aid that they were not happy to proceed with further measurements.

Data collection:

Baseline characteristics were collected from medical notes. GMFCS[13] level was taken from most recent clinic letter and the degree of scoliosis was determined as mild, moderate or severe based on review of previous Paediatric Orthopaedic Clinic letters and/or X-rays when available. Those children who had spinal fusion were classified as having severe scoliosis.

Measurements:

Three health professionals (Measurers) were trained in measuring children according to the method by Haapala et al, 2015[12] for both tibial and recumbent length. We used photographs (in the protocol) and

had two practice sessions with volunteers. One of the Measurers spent two mornings observing the other Measurers at the Special School.

All of the measurements performed at the School for Children with CP were performed by Measurers JM and PC (both doctors), and the majority of the measurements at the Outpatient clinics were performed by Measurers JM and CW (doctor and paediatric dietitian). A small number of children were measured by other trained clinic staff: a physiotherapist, a Rehabilitation nurse and a final year medical student. Unless otherwise stated, the representative and intraobserver measures pesented were made by JM.

All measurements were taken using a steel tape and a washable black marker was used to mark the landmarks. Where possible clothing was removed to allow access to the bony landmark where acceptable. Children had their shoes and ankle-foot-orthoses removed. When it was not acceptable or possible to mark the skin small white dot stickers were used and a mark put on top on the stickers. Stickers and marks were removed between measurements.

Each measurement was taken twice by each Measurer to the nearest 1mm, and if there was more than 2mm difference a third measurement was taken. The average of the two closest measurements taken was used in analysis.

Repeated measures were alternated between measurers: for example measurer one carried out first tibial length measurement, measurer two carried out second tibial length measurement, measurer one carried out third and measurer two the fourth. Each measurer also left the room while the other carried out their measurements.

All measurements were taken on the side of the body that was least affected by CP and if there was no difference then the left side was used.

Acceptability of each measurement was determined by the number of children who declined each measurement. A teacher's aide at the special school was present during both the tibial and recumbent length measuring process. She was able to independently confirm with children that they were happy to commence and to continue with the measurements. A parent was present during the measuring process and after each measurement we checked with both child (where possible) and parent that they were happy to continue with subsequent measurements.

1) Tibial length

Tibial length was measured from the superior border of the medial tibia condyle to the inferior border of the medial malleolus with the knee and ankle at 90 degrees and the child seated.

2) Recumbent length

We used the sum of continuous segmental measures as described in the study by Haapala et al to calculate recumbent length. Thefour measured segments were: (1) from the top of the head to the acromion process of the shoulder, (2) from the acromion process of the shoulder to the greater trochanter of the hip, (3) from the greater trochanter of the hip to the lateral joint line of the knee, and (4) from the knee joint line to the bottom of the heel. This is a novel measurement method and not routinely used in New Zealand (NZ) or internationally.

Landmarks were identified with the children lying supine before measurements were commenced using techniques described in the International Society for the Advancement of Kinanthropometry Manual (ISAK)[14]. The technique was modified by using a headboard with a 90 degree angle.

3) Standing Height

Children without significant contractures or scoliosis who were able to stand straight without assistance had their standing height measured using a stadiometer. They were asked to stand on the centre of the base with their back to the stadiometer and their feet together with heels touching the bottom of the stadiometer[15].

Statistical analysis

Primary data analyses were undertaken using JMP v13.1 (SAS Inc.). The mean difference between measures was tested against the null hypothesis of no difference using the paired t-test.

One way random effects intra-class coefficient (ICC) and Bland Altman 95% limits of agreement(LOA)[16] were calculated using StatsDirect v3. (StatsDirect Ltd).

The technical error of the measurement (TE) and the coefficient of variation (CV) were calculated as described in Bell et al[8]. TE was calculated as $\sqrt{(\Sigma d^2/2n)}$, where d is the difference between the same measure on the same child, and CV is the TE divided by the overall mean of all subjects for the particular measure.

Ethics

Ethical approval for the study was obtained from the Health and Disability Ethics Committee (HDEC) in New Zealand (approval number NZ/1/258A013; 16/CEN/64), the Auckland District Health Boards the Maori Research Committee and the Auckland District Health Board Regional Ethics Committee.

RESULTS

Thirty eight participants consented to be in the study – 23 from the Special School and 19 in the Outpatient Clinic however of those, two children were not measured because they were not at school on the day of measuring, one declined all measurements on the day and one child was excluded

because he did not have CP. The mean age of the study sample was 12 years (range 3-20 years) with 50% female participants. Demographic data are shown in Table 1. (55%) of students at the Special School who were approached to be in the study consented to their child participating.

And (60%) approached in eight outpatient clinics consented to participate.

Table 1: Participant demographic information by GMFCS Level

	All	GMFCS I	GMFCS II	GMFCS III	GMFCS IV	GMFCS V
N	38	6	8	3	5	16
Female %	50	50	63	33	20	56
Ethnicity %						
European	55	67	63	67	60	44
Maori	8	0	0	0	0	19
Pacific	18	17	13	0	40	25
Asian	13	17	13	33	0	6
Other	5	0	13	0	0	6
Scoliosis %	26	0	0	33	20	50
Age (years) mean (SD)	12.4 (5.0)	9.0 (4.0)	12.6 (4.1)	7.6 (5.0)	14.1 (3.4)	14.0 (5.5)

GMFCS=Gross Motor Function Classification

Tibial Length**:**

Seven participants were not able to have intraobserver measurements completed because of time constraints. The intraobserver reliability of tibial length was calculated for each Measurer (Table 2). The

TE for Measurer JM was 0.6cm with a CV of 2.1%. The Bland Altman mean difference was -0.1cm (SD 0.9cm) with 95% limits of agreement (LOA) of -1.9cm and 1.6cm (Figure 1).

Table 2 Intraobserver reliability of tibial length measurement in children with cerebral palsy

Measurer	N	TE (cm)	CV (%)	95% LOA (cm)	ICC
JM	31	0.6	2.1	-1.9 to 1.6	0.98
PC	19	0.7	2.2	-2.8 to 1.8	0.97
CW (and other)	12	0.4	1.5	-1.3 to 1.2	1.00
Combined	62	0.6	2.0	-1.8 to 1.6	0.99

TE=technical error

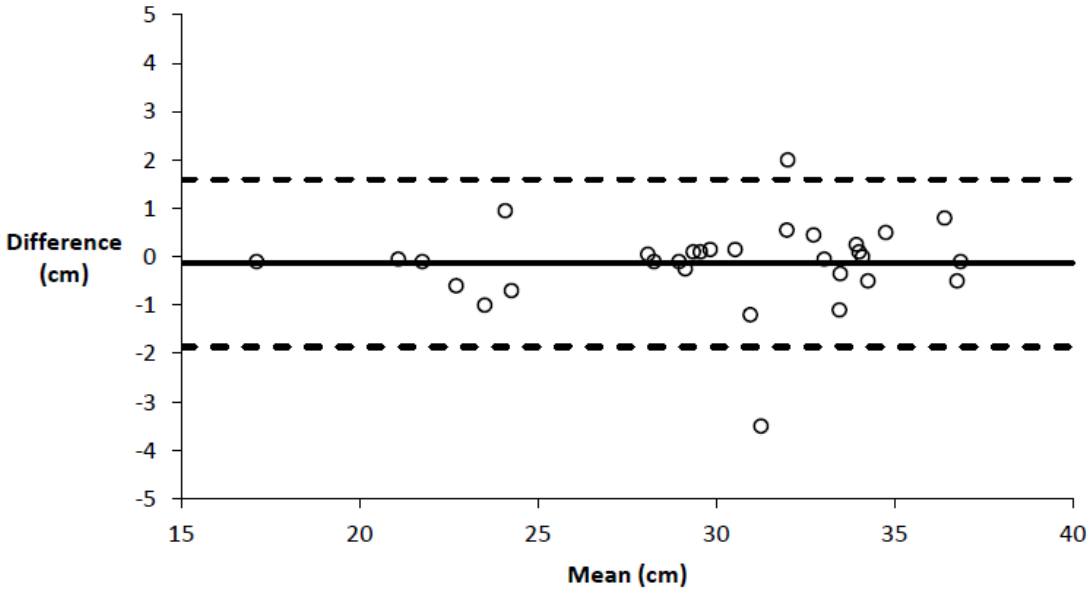
CV=coefficient of variation

LOA=limits of agreement

ICC= intra-class coefficient

Figure 1: Bland-Altman plot for intraobserver reliability of Tibial length (cm) for Measurer JM

Solid line is mean difference. Dashed lines are 95% limits of agreement (LOA)



Three participants did not have interobserver measurements recorded because of time constraints and two had their interobserver measurement disregarded because the protocol was not followed. The interobserver reliability measurements from JM were compared to the combined measurements from PC, CW and one other. The TE was 0.9cm with a CV of 3.1%. The Bland Altman mean difference was 0.4cm (SD 1.3cm) with 95% LOA of -2.2cm and 2.9cm. None of the participants declined any of the tibial measurements.

Recumbent Length:

Intraobserver reliability was measured for 26/38 participants. Seven participants did not have intraobserver recorded for recumbent length because of time constraints; two declined to have any recumbent measurements taken; two declined a second recumbent measurement and one participant declined a third recumbent measurement. The intraobserver reliability of recumbent length was calculated for each Measurer (Table 3). The TE for Measurer JM was 2.0cm with a CV of 1.4% . The Bland Altman mean difference was -0.2cm (SD 2.9cm) with 95% LOA of -5.8cm and 5.4cm (Figure 2).

Table 3 Intraobserver reliability of recumbent length measurement in children with cerebral palsy

Measurer	N	TE (cm)	CV (%)	95% LOA (cm)	ICC
JM	26	2.0	1.4	-5.8 to 5.4	0.99
PC	14	2.6	1.8	-6.5 to 8.8	0.97

CW (and other)	9	1.3	0.9	-2.2 to 4.2	0.99
Total	49	2.1	1.5	-5.7 to 6.1	0.99

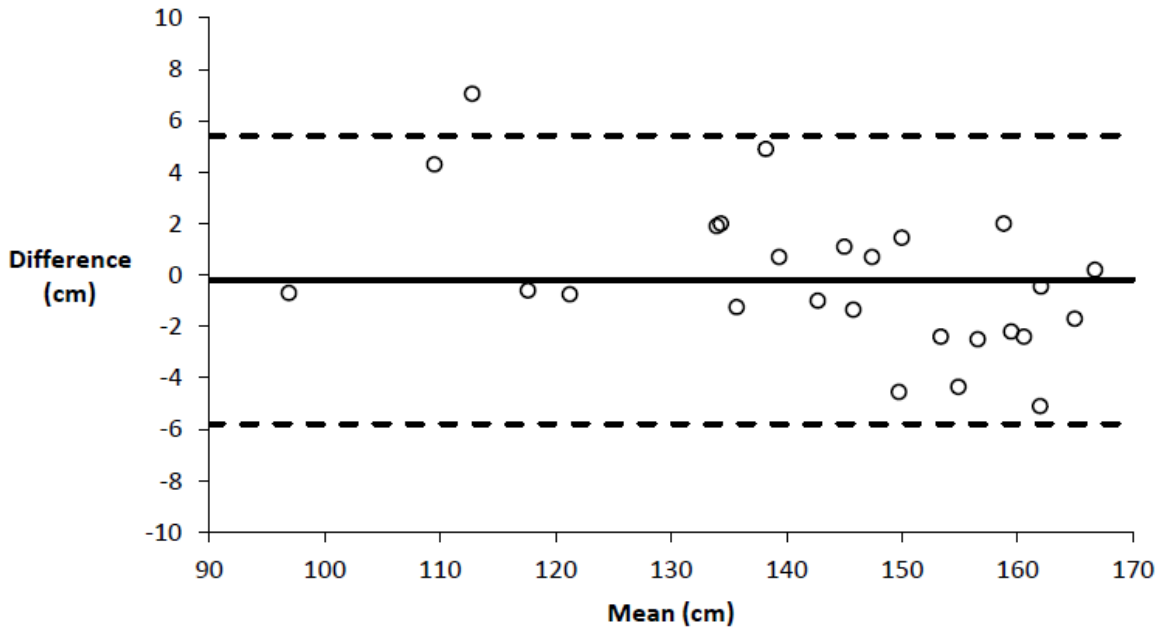
TE=technical error

CV=coefficient of variation

LOA=limits of agreement

ICC= intra-class coefficient

Figure 2: Bland-Altman plot for intraobserver reliability of Recumbent Length (cm) for Measurer JM. Solid line is mean difference. Dashed lines are 95% limits of agreement (LOA).



Three participants did not have interobserver measurements recorded because of time constraints; two participants declined any recumbent measurements to be taken; two declined a second recumbent measurement and one participant declined a third recumbent measurement. For interobserver reliability measuremrents from JM were compared to the combined measurements from PC, CW and

one other. The TE was 2.3cm with a CV of 1.6%. The Bland Altman mean difference was 0cm (SD 3.3cm) with 95% LOA between -6.5cm and 6.5cm.

The intraobserver and interobserver reliability of each segmental length used in the recumbent length measurement are shown in Table 4. Intraobserver reliability was greatest for Segment 4 with TE of 0.6cm and CV of 1.6%. Interobserver reliability was greatest for both segment 1 and 4 with TE of 1.1cm and CV of 4.5% for segment 1 and TE of 1.5cm and CV of 4.1% for segment 4.

Table 4 Intra- and Interobserver reliability of segmental length measurements in children with cerebral palsy

	Intraobserver (N=26)				**Interobserver (N=30)**			
	TE (cm)	CV (%)	95% LOA (cm)	ICC	TE (cm)	CV (%)	95% LOA (cm)	ICC
Segment 1	1.2	5.1	-3.1 to 3.6	0.82	1.1	4.5	-2.9 to 3.1	0.85
Segment 2	1.9	3.8	-5.2 to 5.5	0.94	3.3	6.8	-10.5 to 6.3	0.84
Segment 3	1.6	4.7	-4.9 to 4.0	0.92	2.5	7	-3.7 to 4.1	0.82
Segment 4	0.6	1.6	-1.8 to 1.5	0.99	1.5	4.1	-4.2 to 4.1	0.94

TE=technical error

CV=coefficient of variation

LOA=limits of agreement

ICC= intra-class coefficient

Agreement between Standing Height and Segmental Recumbent Length:

For fourteen participants with GMFCS I-III who were able to stand straight, both standing height and recumbent length using continuous segmental measures were recorded (57% females; mean age 9 years). The Bland Altman mean difference was 3.7cm (SD 3.2cm, $p<0.001$) with 95% LOA of -2.5cm and 10.0cm.

DISCUSSION

We found that the tibial measurement was convenient, quickly undertaken and acceptable to children with CP. No children or young people refused to have tibial measurements done. Unfortunately however our measurements for both interobserver and intraobserver were not as reliable as in other comparable studies. For example for Measurer JM the TE for tibial length intraobserver was 0.6cm and the CV was 2.1%; the interobserver TE was 0.9cm and CV 3.1%, compared to Spender et al[7] where the TE for intraobserver was 0.2cm, the CV was 0.84%; the interobserver TE was 0.49cm and CV was 2%.

Our tibial length measurements may have been less reliable compared to other studies because of our short training period and the time pressure in clinic. The tibial length measurements by CW and other were the closest in reliability to other studies[3,7]. CW has had more experience in using this measurement in her clinical practice and this may explain the higher reliability.

Our segmental length measurements were also less reliable than those in Haapala et al[12]. The difficulties faced when trying to measure segmental length were greater than when measuring tibial length. The child or young person had to be moved to lie supine and once on the bed they usually took time to settle. This may have influenced the results as the child sometimes moved position during measurement. The study took place over winter and the children were heavily clothed so it was difficult to expose the bony landmarks. Some measurements were taken over clothes which may have moved during the process and therefore influenced the reliability of the measurement also. Overall it took much longer to measure recumbent compared to tibial length.

According to Ulijaszek et al, 1999[17] a “good” measurement error for height/length in typically developing children is less than 0.5cm with “gross error” being greater than or equal to 2cm. This

indicated our error for recumbent length was unacceptable. Unlike Haapala et al[12] our recumbent length measurement also did not accurately approximate standing height.

In regards to acceptability, all subjects allowed tibial length measurements to take place, however some participants refused to have segmental length measured, or re-measured. This indicated to us us that segmental length as described in our study is not a feasible measurement to advocate for use across New Zealand, even if the training and thus perhaps measurement error could be improved.

Strength and Limitations of the Study

This is the first study to investigate measuring tibial and recumbent length in a sample of New Zealand children with CP. A strength of this study was that some of the measurements were carried out in a clinic setting which reflects the reality of where children with CP will most often be measured. We also used standard methods for measurement, and measurers were trained in these techniques. We included participants with moderate to severe CP and felt confident assessing the acceptability of each of measurements despite difficulties with communication because of the assistance of a teacher's aide at the special school.

A limitation was that it was difficult to recruit patients in the clinic setting because parents had often not received their participant information in the mail prior to the appointment and had not allowed any extra time to have measurements done. This study had a small sample size, which varied widely in a number of factors including age, degree of scoliosis and severity of CP. The study size was too small to conduct reliable subanalyses to determine if the reliability of the specific method varied by age and severity of CP. Due to cross sectional nature of the study we were also unable to determine the validity of the measurements over time.

Conclusion

We suggest that tibial length could be recommended for measuring the growth of children with moderate to severe CP. It is easy to measure in the Outpatient setting, therefore feasible and acceptable by children. It can be plotted directly onto tibial length growth charts. Further standardisation of methods and training however is required to improve measurement accuracy and reliability.

Longitudinal studies with repeated measures are needed to determine if either measurement is suitable to monitor growth over time.

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protocol and we would also like to thank the researchers of the Haapala et al study for clarifying via email correspondence the landmarks used in their study protocol. Lastly but most importantly we would also like to thank the staff at both the Carlson Special School and the Wilson Centre in Auckland for their support and all the children and their families who participated in the study.

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