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博　士　論　文

Relationship between stature and tibial length for children with moderate-to-severe cerebral palsy

（中等度から重度の脳性麻痺児における身長と脛骨長の関係）

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Relationship between stature and tibial length for children with moderate-to-severe cerebral palsy

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Abstract

Objective:
To derive the equation for estimating stature, based on tibial length, for children with moderate-to-severe cerebral palsy (CP) and lower limb joint contracture or scoliosis.

Methods:
The participants (3–12-years-old) included 50 children with moderate-to-severe CP (mean age, 8.3 ± 2.4 years) and 38 typically developed (TD) children (mean age, 7.5 ± 2.6 years). Thirty-four (68%) of the children with CP had a gross motor function classification system level of V. Furthermore, 40 (80%) had definite lower limb joint contracture or scoliosis. The stature and the tibial length measurements of all participants were determined. Regression equations to estimate stature, based on tibial lengths, were determined for both TD children and children with CP. Moreover, regression equations defining the relationship between tibial length and age were compared between the two groups of children, using multiple regression analysis.

Results:
The regression equations for estimating stature, based on tibial length, were stature = tibial length × 3.25 + 34.45 [cm], $R^2 = 0.91$ (TD children), and stature = tibial length × 3.42 + 31.82 [cm], $R^2 = 0.81$ (CP children). In children with CP, tibial lengths were significantly shorter than those in similarly aged TD children.

Conclusion:
The stature of children with moderate-to-severe CP can be estimated from their tibial lengths, regardless of the presence of joint contracture or scoliosis. The tibial length may be a proxy for estimating stature during the growth assessment of children with moderate-to-severe CP.

Keywords: cerebral palsy; stature; measurement; tibial length; reliability

Introduction

The growth and nutritional assessments of neurologically impaired children is important because they often demonstrate growth failure and malnutrition [1, 2, 3, 4, 5]. Stature and weight measurements are fundamental anthropometric indices of growth assessment. Previous studies reported that children with moderate-to-severe cerebral palsy (CP) tend to have lower statures and weights than similarly aged, typically developed (TD) children [3, 4, 6, 7]. Stevenson et al. [3], reported that children with CP, tended to have worse health indices and less social participation (e.g., school or usual activities). Unfortunately, among children with moderate-to-severe CP, accurate stature measurements are difficult because most cannot stand without support and they often have joint contracture and scoliosis [3, 4, 8, 9].

As part of the growth and nutritional assessments of children with CP, both stature and weight values should be obtained. Although weight measurements are easy and reliable, the methods to measure stature, particularly for children with moderate-to-severe CP, have not been established. However, several methods have been reported to estimate the stature of persons who cannot stand. Stature can be accurately estimated from knee height [9, 10, 11, 12] or long bone length [4, 9, 13]. Of these methods, the estimation of stature from long bone length might be useful for children with moderate-to-severe CP who also have joint contracture and scoliosis. Similarly, stature estimation tibial length, was reported to have good accuracy in children with CP who did not have joint contracture or scoliosis [4, 9]. However, the relationship between tibial length and the stature of children with moderate-to-severe CP and lower limb joint contracture or scoliosis is difficult. Furthermore, there have been few reports regarding tibial growth in children with moderate-to-severe CP.

Previously, we reported the reliability of stature and tibial length measurements of children with moderate-to-severe CP [14]. The methods demonstrated good intra- and inter-rater reliability.

The purposes of this study were to (1) determine the correlation between reliable stature determinations and tibial lengths in children with moderate-to-severe CP, (2) examine the validity and usefulness of a regression equation to estimate stature based on tibial length for these children, and (3) compare tibial growth between children with moderate-to severe CP and TD children.

Participants and methods

Participants

Fifty children (3–12-years-old) with CP and 38 TD children were enrolled in this study. The children with CP were classified according to the gross motor function classification system (GMFCS) [15]; the presence of lower limb joint contracture and scoliosis was determined from a medical records review. Joint contracture was defined as an abnormal and permanent condition of a joint, due to shortening of the muscle tissue. In this study, participants who were unable to position their hip and knee joints at 0° extension were classified as "with joint contracture". The presence of scoliosis was diagnosed by pediatric orthopedic physicians based on X-ray films. Furthermore, the estimated onset of injury for each child with CP was also determined from the medical record review. All participants with CP were treated at the same medical center, either in the outpatient clinic or after admission, between 2010 and 2014. Children with histories of growth hormone treatment or bone disorder complications were excluded. The TD children were recruited using a snowball sampling method from the regions where the participants with CP lived. Children with physical or intellectual problems noted during their usual health check-ups were excluded from the TD participants group. To maintain ethnic uniformity, only children with Japanese parents were included.

Ethics

This study was approved by the Ethics Committee of Kobe University Graduate School of Medicine in accordance with the Helsinki Convention. Participants were provided information according to their ability to understand, and all participants (or their parents) agreed with the study aims.

Measurements

For the TD participants, tibial lengths were measured from the superomedial edge of the tibia to the inferior edge of the medial malleolus, using a tape measure [9, 14]. Statures were determined using each individual's standing height. For participants with CP, stature was measured using a tape measure with the patient in a prone or side-lying position, using a division-method [14]. In the division-method, the stature is defined as the sum of the distances from the plane contacting the vertex to the spinous process of the seventh cervical vertebra; the length from the spinous process of seventh cervical vertebra to the Jacoby line, along the spinal curvature; and the length from the Jacoby line to the lower rim of calcaneus bone along the posterior aspect of the lower limb. All measurements were determined to the nearest 0.1 cm. All measurements, except for the stature of TD children, were determined twice, and the average was recorded. The stature of TD children was measured once.

Statistical analysis

The regression equation used to estimate stature from the tibial length was calculated using the least squares method for both TD children and children with CP. The regression equations for the two groups of children were compared using a multiple regression analysis. In this analysis, the tibial length of participant and attribute (TD = 0, CP = 1) were defined as the independent variables and the stature was defined as the dependent variable. Furthermore, the correlation between individual age and tibial length was calculated using the least squares method. The regression equations for the two groups of children were compared using a multiple regression analysis. In this analysis, the participant age and attribute (TD = 0, CP = 1) were defined as the independent variables and the tibial length was defined as the dependent variable. The statistical analyses were performed using IBM SPSS Statistics for Windows (version 19; IBM, Armonk, NY, USA) and Excel for Windows (version 14.0; Microsoft, Redmond, WA, USA).

Results

Participant demographic characteristics

Table 1 shows the participant demographic characteristics. The average age of the children with CP (mean ± SD) was 8.3 ± 2.4 years, 54% were male. The GMFCS classification of the children with CP indicated that the majority (34, 68%) were level V. Forty (80%) of the children with CP had definite lower limb joint contracture or scoliosis, with most (30, 60%) estimated to have developed the injury during the perinatal period. The average age of the TD children was 7.5 ± 2.6 years, 61% were male. There were no significant differences between the TD children and children with CP with respect to their average age (Mann-Whitney U-test, $p = 0.18$) or their sex distribution (χ^2 test, $p = 0.54$).

Regression equation relating stature and tibial length in TD children and in children with CP

Figure 1 and Table 2 show the regression equation describing the relationship between stature and tibial length for the 38 TD children. Figure 2 and Table 2 show the regression equation describing the relationship between stature and tibial length for the 50 children with CP. The regression obtained values for the children with CP and the TD children were similar.

Table 3 shows the results of a multiple regression analysis of the relationship between stature and tibial length for both groups of children. The multiple regression equation was:as follows

$$\text{Stature} = 3.25\ (x_1) - 2.63\ (x_2) + 0.17\ (x_1x_2) + 34.45\ [\text{cm}]\ (1).$$

The coefficient of determination of the regression equation (R^2) was 0.86. In the equation, x_1 and x_2 were independent variables: x_1 is the child's tibial length, x_2 is the child's attribute (TD = 0, CP = 1), and x_1x_2 signifies the interaction. The 95% confidence intervals (CI) for the partial regression coefficients of x_2 and x_1x_2 were 0. This relationship means that, there was no significant differences between the regression equations for TD children and for children with CP.

Relationship between tibial length and age for the TD children and children with CP

Figure 3 shows the regression equations describing the relationship between tibial length and age for both the TD children and children with CP. For both groups of children, there were good correlations between tibial length and age. For children with CP, the slope and the intercept of the regression were smaller than those for TD children. The R^2 of the regression equation for children with CP was also inferior to that for TD children.

Table 4 shows the results of a multiple regression analysis of the relationship between tibial length and age for both groups of children. The multiple regression equation was:as follows

$$\text{Tibial length} = 1.74\ (x_1) - 0.82\ (x_2) - 0.43\ (x_1x_2) + 13.37\ [\text{cm}]\ (2).$$

Here, x_1 and x_2 were independent variables: x_1 is the child's age, x_2 is the participant's attribute (TD = 0, CP = 1), and x_1x_2 signifies the interaction. The partial regression coefficient of x_1 and its 95% CI were obvious for values >0. On the other hand, the partial regression coefficient of x_1x_2 and its 95% CI were obvious for values <0. This relationship indicates that, for both TD children and children with CP, tibial lengths increased as the participants aged. However, the rate of tibial lengthening was lower for children with CP than for TD children, with an R^2 of 0.77.

Discussion

In this study, regression equations for the relationships between stature and tibial length were obtained for both TD children and those with moderate-to-severe CP. The

R^2 values for both regression equations were good. The regression value determined for TD children was similar to that reported in a previous study using X-ray photography of the tibia in TD children [16]. For the children with moderate-to-severe CP, the regression value obtained was similar to that obtained for children with CP who had no joint contracture or scoliosis [4, 9]. Furthermore, the obtained regression values for the children with CP and TD children were similar.

Previous studies have reported other methods for estimating the stature of children with CP. For those with joint contracture or scoliosis, recumbent body length has been measured [1, 6, 8]. However, the methods have not been reported with definite procedures; therefore, the measurement reliabilities might be poor [6, 8]. The method for estimating stature based on knee height is popular [9, 10, 11, 12]; nevertheless, knee height cannot be measured in children with CP who have severe knee and ankle joint contracture. Although, the method to estimate stature using tibial length has been previously reported [4, 9], only children with CP not having joint contracture or scoliosis were included in the previous study.

In the present study, a strong correlation was found between tibial length and stature in children with moderate-to-severe CP and lower limb contracture or scoliosis. The derived regression equation was similar to that for TD children and children with CP who had no joint contracture or scoliosis. Our original supposition that children with moderate-to severe CP, regardless of lower limb joint contracture or scoliosis, would have tibial lengths, as a proportion of stature, similar to those for TD children was shown to be accurate. Further, our results suggest that the stature of a child with moderate-to-severe CP might be estimated using tibial length regardless of the presence of lower limb joint contracture or scoliosis. In the division-method, although the spine length might be overestimated when an individual had kyphoscoliosis [4], the measurement can be performed reliably. However, the measurement requires specialized training. Furthermore, this method is time-consuming because of the complicated measurements (i.e., children must be prone or lying on their sides). In contrast, tibial length measurements can be completed easily. Moreover, the correlation between stature and tibial length has been reported to be equal to or better than those of between stature and other long bones or the spine in children with CP who do not have joint contracture or scoliosis [4]. Thus, tibial length is a reliable index of bone growth and may be a proxy for stature for use in growth assessments of children with moderate-to-severe CP.

Kong et al. [4] reported that the bone age of children with CP was delayed in comparison with that of TD children. Our study showed that tibial length increases, in children with moderate-to-severe CP, were delayed compared with similarly aged TD children. Furthermore, in children with CP, the scatter of tibial lengths according to age was wider than for TD children. In children with CP, previous studies have also indicated that the long bones lengthen at a slower rate than do those of TD children [4, 13]. Such studies have suggested that malnutrition, growth hormone deficiencies, or bone loading might affect bone growth. However, the factors responsible for poor bone growth remain unclear. Further investigation of the factors affecting bone growth in children with CP is needed.

This study has several limitations. The factors influencing tibial length in children with CP were not studied. The number of subjects was relatively small. In addition, all of the participants with CP were recruited from one facility, and the data were obtained solely from Japanese children. Thus, our regression equation may not apply to non-Japanese ethnic groups. The inclusion of additional participants may elucidate additional factors that influence the regression equation relating stature and tibial length in children with CP. Further investigations are required.

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Figure Legends

Figure 1. The regression equation describing the relationship between stature and tibial length for 38 typically developed children, aged 3–12 years.
R^2 : The coefficient of determination of the regression equation.

Figure 2. The regression equation describing the relationship between stature and tibial length for 50 children with moderate-to-severe cerebral palsy, aged 3–12 years.
R^2 : The coefficient of determination of the regression equation.

Figure 3. The regression equation describing the relationship between tibial length and age for 38 typically developed children and 50 children with moderate-to-severe cerebral palsy, aged 3–12 years.
R^2 : The coefficient of determination of the regression equation.

Table Legends

Table 1. GMFCS, gross motor function classification system.

Table 2. TD, typically developed; CP, cerebral palsy; SE, standard error; CI, confidence interval.
The stature of TD children determined using their standing height; CP children measured using the segmental method.

Table 3. TD, typically developed; CP, cerebral palsy; SE, standard error; CI, confidence interval.

Table 4. TD, typically developed; CP, cerebral palsy; SE, standard error; CI, confidence interval.

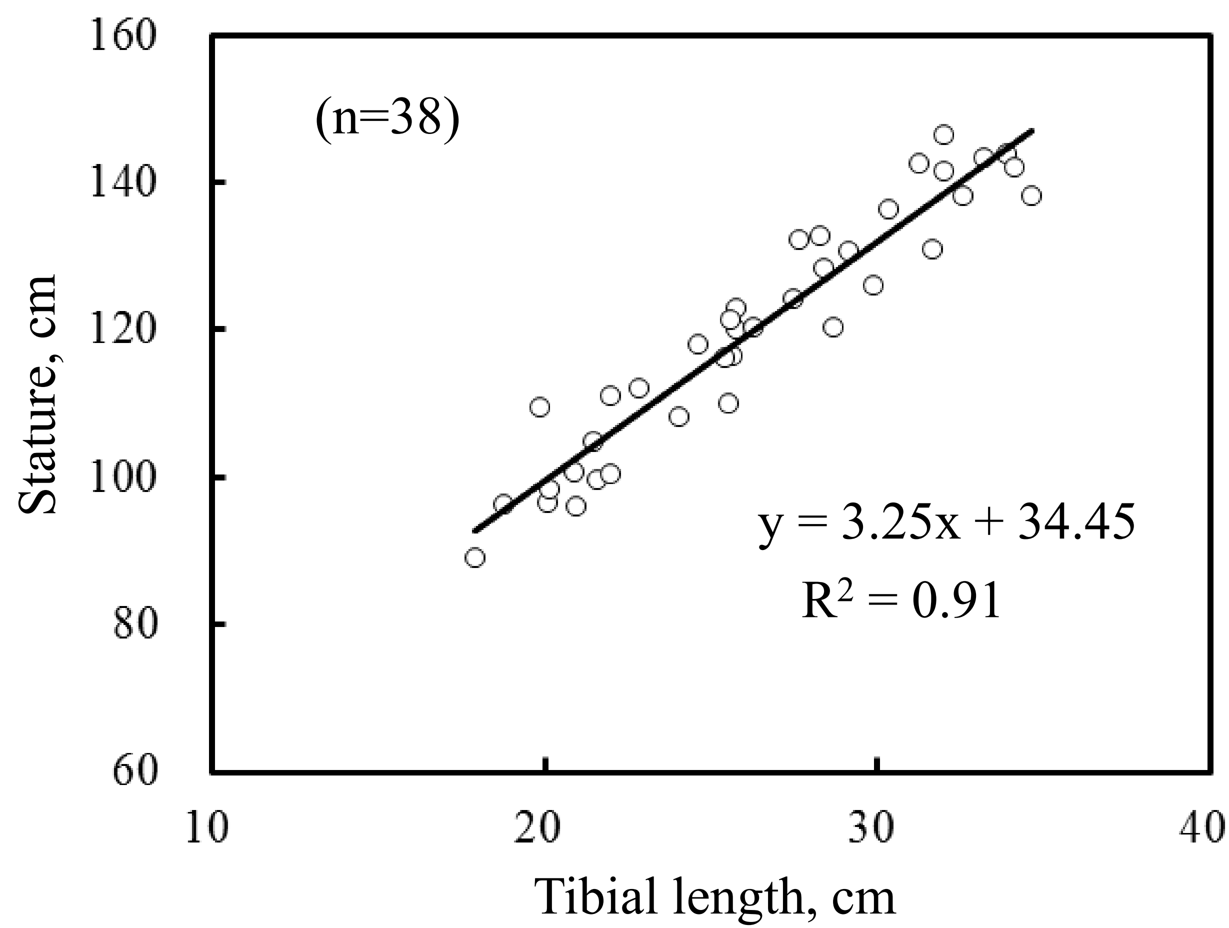

Fig. 1 – The regression equation describing the relationship between stature and tibia length for 38 typically developed children, aged 3-12 years.

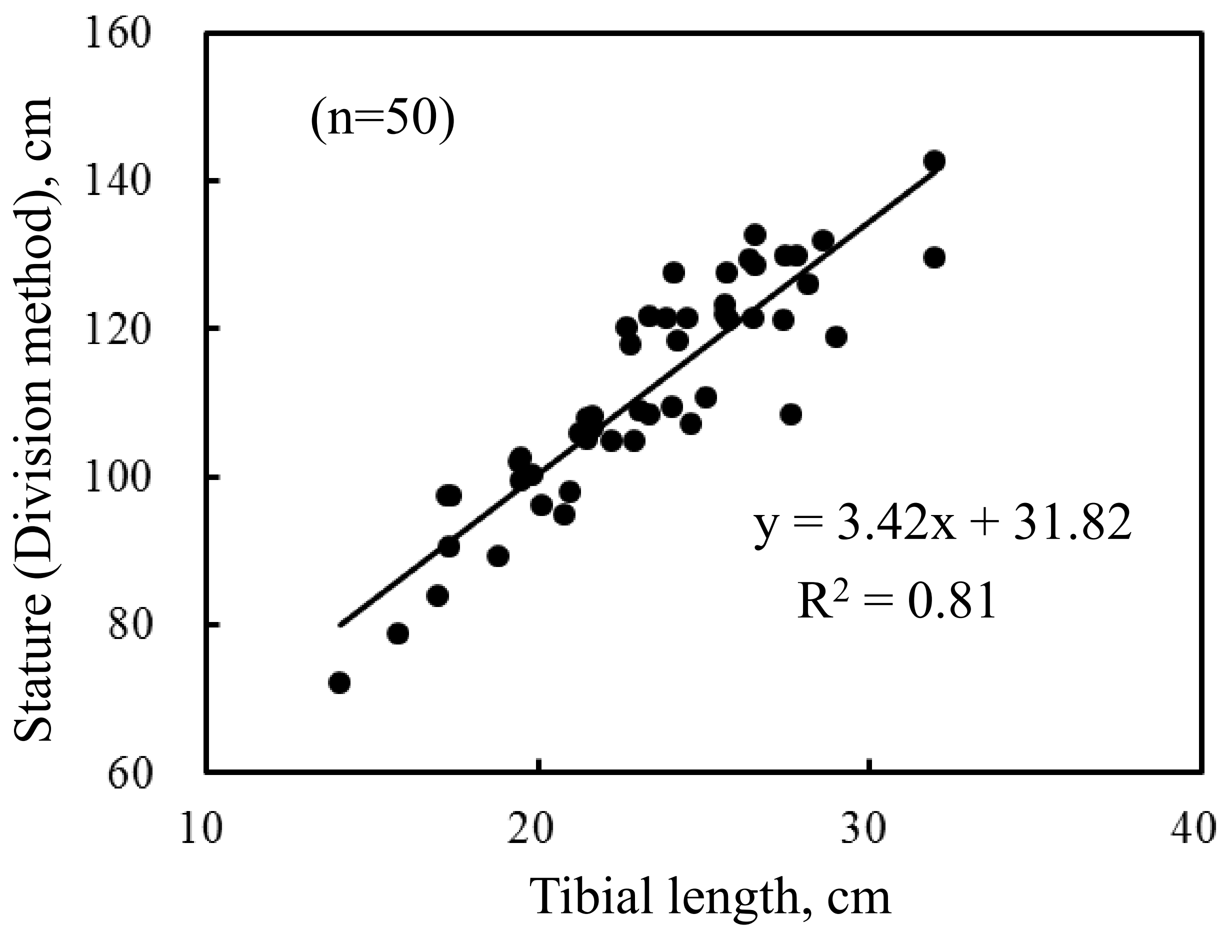

Fig. 2 – The regression equation describing the relationship between stature and tibia length for 50 children with moderate-to-severe cerebral palsy, aged 3-12 years.

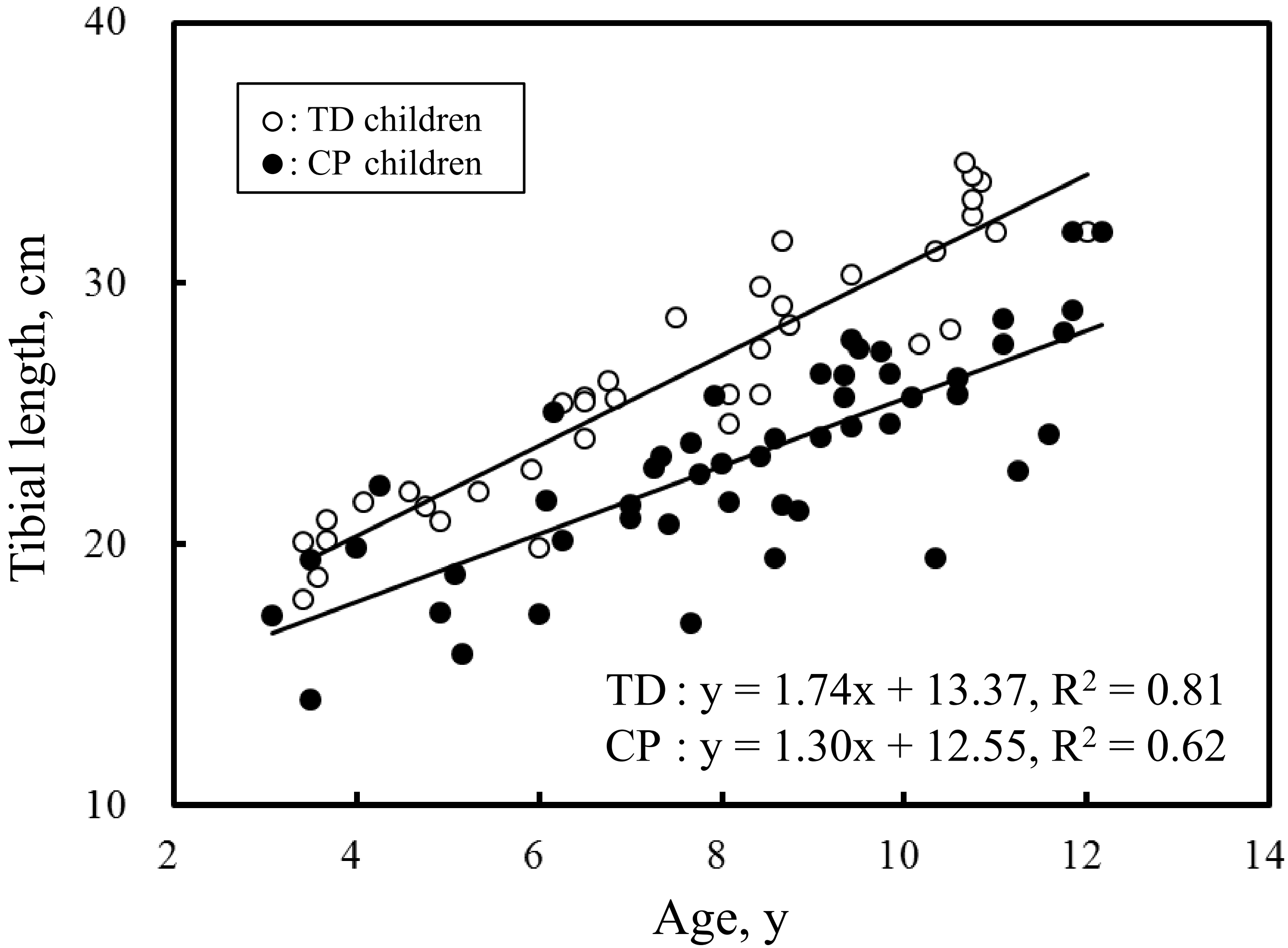

Fig. 3 – The regression equation describing the relationship tibia length and age for 38 typically developed children and 50 children with moderate-to-severe cerebral palsy, aged 3-12 years.

Table 1

Demographic characteristics of the participants with cerebral palsy

	N
GMFCS classification	
Level III	8
Level IV	8
Level V	34
Existence of scoliosis	
with scoliosis	24
without scoliosis	4
have not examined	22
Existence of joint contracture of lower limb	
with contracture	38
without contracture	12
Estimated period of injured	
prenatal	11
perinatal	30
post natal	9

Table 2

Regression equation of stature on tibia length for TD children and children with CP

	Equation	SE [cm]	R^2	N	Mean of stature* [cm]	Mean of tibia length	SE and 95%CI, Slope	SE and 95%CI, Intercept
TD children	3.25(x)+34.45	4.91	0.91	38	120.19±16.38	26.38±4.81	0.17, [2.91, 3.59]	4.50, [25.33, 43.57]
CP children	3.42(x)+31.82	6.69	0.81	50	111.59±15.21	23.31±4.00	0.24, [2.94, 3.90]	5.64, [20.47, 43.17]

Table 3

Partial regression coefficient between stature and tibial length for TD children and children with CP

	Partial regression coefficient	SE and 95%CI	p-value
Tibial length (x_1)	3.25	0.20, [2.84, 3.66]	<0.001
Attribute (x_2) (TD = 0, CP = 1)	-2.63	7.46, [-17.47, 12.21]	0.73
Tibial length × Attribute (x_1x_2)	0.17	0.30, [-0.42, 0.76]	0.57
Intercept	34.45	5.49, [23.54, 45.36]	<0.001

Table 4

Partial regression coefficient between tibia length and age for TD children and children with CP

	Partial regression coefficient	SE and 95%CI	p-value
Age (x_1)	1.74	0.14, [1.46, 2.01]	<0.001
Attribute (x_2) (TD = 0, CP = 1)	-0.82	1.58, [-3.96, 2.33]	0.61
Age × Attribute (x_1x_2)	-0.43	0.19, [-0.81, -0.05]	0.03
Intercept	13.37	1.11, [11.16, 15.58]	<0.001