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Lower Extremity Strength and Hopping and Jumping Ground Reaction forces in Children with Neurofibromatosis Type 1

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Abstract

The purpose of this study was to 1) extend the research findings of decreased muscular force production in grip strength to the lower extremity strength of children with NF1 and 2) to determine if there was a relationship between isometric strength and functional activities in children with NF1. Force production was assessed using a hand held dynamometer (HHD) and a functional task (hopping and jumping) on a force plate. Data from twenty-six children with NF1 were compared to data from 48 typically developing children of similar sex, weight and height. Children with NF1 demonstrated statistically significant lower force production with HHD ($p < 0.01$) during hip extension, but similar force production for knee extension and ankle plantar flexion compared to the control group. A relationship existed between the ground reaction forces at take-off from both hopping and jumping and the force generated from knee extensor strength in the NF1 group. The addition of a functional task to hand held dynamometry is useful for determining a relationship between common clinical measures and functional activities.

Keywords

Neurofibromatosis type 1; strength; muscle force; hand held dynamometry; jumping/hopping peak ground reaction forces

1 Introduction

Neurofibromatosis type 1 (NF1) is a common autosomal dominant disorder presenting in childhood. Examples of the clinical manifestations of NF1 include café-au-lait macules, tumors of peripheral nerves, optic pathway tumors, long bone dysplasia, developmental delays, and learning disabilities. NF1 is associated with skeletal abnormalities such as short stature, scoliosis, and long bone fracture with non-union. Children with NF1 have

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abnormalities of bone and muscle as evidenced by decreased bone mineral density, decreased bone strength, and low muscle mass, all of which may predispose them to fractures and scoliosis (Stevenson, Murray, Viskochil, Carey, & Moyer-Mileur, 2008; Tucker et al., 2009). Weakness in hand grip strength has been documented in children and adults with NF1 (Souza, Passos, Guedes, Rezende, & Rodrigues, 2009). Souza et al. (2009) concluded that reduced muscle force may be an impairment associated with NF1.

Beenacker, van der Hoeven, Fock, and Maurits (2001) suggested the maximum isometric contraction values obtained from hand held dynamometry testing in children could be influenced by testing position, technique, and the opposing strength of the examiner making it necessary to account for confounding variables when using dynamometry testing as a measure of strength. Testing position and technique could be standardized to account for variability between examiners; however, it is difficult to control for examiner strength between studies and between clinicians. Rose et al. (2008) used leg length to compute torque values in a group of 6 to 8 year olds to adjust for distance to the joint center for children of different heights. Using torque measures can adjust for height differences, but does not account for sex and weight differences. Additionally, most functional movements do not involve isometric contraction in a fixed position and measuring muscle capability with a functional task may provide a better approximation of muscle strength (Deusinger, 1984). Cordova and Armstrong (1996) found that peak ground reaction force at take off measured during a one-legged vertical jump test was a reliable measure of peak vertical force and that quantifying this force may be an accurate measure of functional lower extremity strength in adults.

A variety of methods exist for documenting changes in jump performance using force plates. Calculations of peak power, peak force, and peak velocity are commonly used to quantify changes in physiological adaptations to strengthening which reflect peak performance improvement. Cormie, McBride, and McCaulley (2009) described the use of power, force and velocity time curves that were able to identify adaptations to jump technique contributing to performance improvement. Hori et al. (2009) reported high reliability of peak power, peak force, and peak velocity measurements derived from ground reaction forces for assessing leg extensor strength ($ICC = .92-.98$). For this study, we were not interested in measuring jump performance, but evaluating the feasibility of using a functional task to represent strength and its relationship to clinical measures of strength. Both hand held dynamometry and peak ground reaction forces are direct measures of net external muscle force production, providing a comparable method of evaluating the force generated by muscles during both a static and dynamic task.

The purpose of this study was to (1) determine if the findings of decreased muscular force production could be replicated in a cohort of children with NF1 in lower extremity muscles using hand held dynamometry and (2) to determine if there was a relationship between force production on hand held dynamometry (isometric strength) and peak ground reaction force at take off in jumping and hopping (functional strength) in children with NF1.

2. Methods

A quasi-experimental study was designed to evaluate force production and functional strength of lower extremity muscles of children with NF1. Typically developing children were recruited from friends and family members of Shriners Hospitals for Children employees. Children with NF1 were recruited from the University of Utah NF1 clinic and through advertising with a local NF1 support group organized by the Children's Tumor Foundation. Participants were examined by one investigator (DS) to confirm the clinical diagnosis of NF1. Only individuals who fulfilled the NIH clinical diagnostic criteria for NF1

were included (Gutmann et al., 1997, 1988). Exclusion criteria were the presence of an orthopedic procedure within the last six months, or tibial dysplasia. These conditions were excluded to reduce the influence of surgery or a musculoskeletal impairment on lower extremity strength. Institutional Review Board approval from the University of Utah was obtained for two separate studies (one to study children with NF1, and one for dynamometry data collection in typically developing children). All children who agreed to participate and met the inclusion criteria were enrolled in the study and completed the evaluation.

2.1 Instruments

Strength was assessed at the Shriners Hospitals for Children Salt Lake City Movement Analysis Laboratory by two experienced physical therapists (BJ & AS). Lower extremity strength was tested in all individuals using a handheld dynamometer (PowerTrackII, JTech, Salt Lake City, UT). Standardized test protocols were used for positioning of the patient, dynamometer placement, and stabilization of proximal joints or limbs (Table 2). One therapist's technique was chosen as the "gold standard" and video taped. The second therapist watched the training video, and feedback was given during practice sessions. The dynamometer was placed perpendicular to the segment being tested. A "make test" was employed in which the examiner "meets" the resistance the participant produces and attempts to prevent the joint from moving. The participant is encouraged to produce their maximum effort and asked to push against the dynamometer for three seconds. The highest force value of four repetitions (two from each limb) of each muscle group was selected for strength analysis. Hip extension, knee extension, and ankle plantar flexion strengths were recorded using the dynamometer. These muscles were chosen to reflect primary muscle groups contributing to power output during jumping and hopping (Nagano, Komura, Fukushima, & Himeno, 2005). The female participants in the NF1 group were significantly smaller in height and weight than the female participants in the control group. The strength values for the control group were derived from computing a value based on the sex, height, and weight of the participants in the NF1 group. A regression equation (Macwilliams, Shuckra, & Mavor, 2010) was used to compute the predicted strength values in order to match the participants between the two groups accurately. The control data were collected by the same clinicians in our laboratory using the same methodology from participants recruited from the same geographical area.

Jumping and hopping were assessed on a force plate (AMTI OR-6, Watertown, MA, USA) integrated in the floor of a laboratory. Data was sampled at 1000 Hz and filtered with a 10 Hz fourth order Butterworth filter. Participants were asked to stand in the middle of the force plate. A maximal effort and technique for jumping and hopping were demonstrated by the therapist and children practiced prior to data collection. Data collection was begun when the child indicated they were ready. Jump data was collected first for all participants, followed by dominant leg hopping, and then non-dominant leg hopping. The dominant leg was determined using the protocol from the Bruininks Oseretsky Test of Motor Proficiency 2 (the leg used for kicking). For jumping, data capture was begun and then participants were asked to "jump as high as you can and land in the same place". For hopping, data collection was begun while participants were standing on both feet. Participants were instructed to pick up one foot and to hop as high as they could and land in the same place on one foot. Participants were allowed to use their arms when jumping and hopping. Since our participants were young children with variable performance, five trials were collected for jumping, hopping on the dominant foot, and hopping on the non-dominant foot to assure collection of 2 useable trials. The data was processed using Vicon Nexus software (Vicon Motion Systems, Centennial, CO). The highest value for peak vertical ground reaction force at take off was normalized by body weight and used for data analysis.

2.2 Data analysis

Post hoc power analysis was performed using an effect size of .99, a sample size of 26, and with alpha set to .05 resulting in a power of .95 (G*power 3, Heinrich-Heine University, Düsseldorf, Germany). The data were analyzed using SPSS software version 14.0 (Chicago, IL). All data were shown to be normally distributed by QQ plot analysis. Descriptive statistics were used to report subject characteristics (Table 1). A one-way ANOVA was used to assess differences between and within groups. Independent student t-tests were used to compare the mean strength values of the NF1 group to the mean strength values of the control group and to evaluate the differences between limbs and between hopping and jumping peak ground reaction forces. General linear regression was used to evaluate the relationship between force production during hand held dynamometry of the hip extensors, knee extensors, and ankle plantar flexors and raw peak vertical ground reaction forces during jumping and hopping.

3. Results

Twenty-six children with NF1 participated in the study (Table I). The control group and NF1 group were similar in age and BMI. The female participants in the NF1 group were significantly smaller than the female and male control subjects in height (18.6 cm, $p < .01$), and weight (10 kg, $p < .03$). The force generated by hand held dynamometry of three lower extremity motions (hip extension, knee extension, and ankle plantar flexion) from the NF1 group was compared to a derived force value from 48 control participants to account for the size differences (Table 3). The NF1 group demonstrated statistically significant lower mean force values in hip extensors ($t = -3.57$, $p < .01$). There were no statistically significant differences in knee extensor force values ($t = -.42$, $p = .67$) and ankle plantar flexor force values ($t = -.65$, $p = .52$) between the NF1 group and the control group.

There was not a statistically significant difference between the peak vertical ground reaction forces at take off between dominant and non-dominant legs during hopping ($t = .43$, $p = .67$). There was not a statistically significant difference between peak vertical ground reaction force at take off for hopping and peak vertical ground reaction force at take off for jumping ($t = 1.39$, $p = .17$). (Table 4) There was a highly correlated ($R = .86$) and statistically significant relationship between peak vertical ground reaction forces at take off in jumping and knee extensor force ($\beta = .698$, $p < .01$) and peak vertical ground reaction forces at take off ($R = .87$) in hopping and knee extensor force ($\beta = .855$, $p < .01$). Knee extensor force accounted for 50%, and 71% of the variance respectively. There was not a statistically significant difference between jumping and hip extensor force ($\beta = .148$, $p = .28$) or ankle plantar flexor force ($\beta = .598$, $p = .56$) or between hopping and hip extensor force ($\beta = -.161$, $p = .32$) or ankle plantar flexor force ($\beta = .121$, $p = .57$). (Table 5)

4. Discussion

The results of this study indicate that children with NF1 have significantly less force production in hip extensor muscles than the control group. However, there was not a statistically significant difference in force production of knee extensors or ankle plantar flexors between the children with NF1 and the control group. Souza et al. (2009) also found decreased muscular force production in the hand grip of adults and children with NF1. These two studies provide evidence that there is an underlying strength and power deficit for individuals with NF1 for some muscles. However, several questions remain to be answered. It is not known whether there is a greater decline in force with age in individuals with NF1 since Souza's study included both adults and children, or if there is a pattern of decreased muscular force with some muscles being more affected than others. It is not known whether

the variation in muscular strength is a primary impairment of NF1 or if it is a secondary impairment attributed to decreased physical activity or low motor competence.

The decreased force production in the hip extensor muscles in NF1 may have clinical consequences. There is a strong relationship between lean body mass and leg power to bone mineral density (Witzke & Snow, 1999). Decreased BMD has been reported in children with NF1 (Stevenson, Murray, Viskochil, Carey, & Moyer-Mileur, 2008) and one of the regions with the lowest BMD scores in NF1 has been reported to be the trunk (Kuorilehto et al., 2005; Yilmaz, Ozmen, Bora, Goksan, & Eskiurt, 2007). Scoliosis, both the dystrophic and non-dystrophic form, is seen in approximately 10-33% of individuals with NF1 (Vitale, Guha, & Skaggs, 2002; Illes, Halmai, de Jonge, & Dubousset, 2001). Illes et al. (2002) reported significant decreases in lumbar BMD in individuals with NF1 who had scoliosis, and there was a correlation of low BMD with the degree of curvature. It is possible that proximal weakness in individuals with NF1 contributes to the development of regional osteopenia and scoliosis commonly observed in NF1. Additionally, hip extensor weakness may contribute to the decreased motor proficiency identified in children with NF1 (Johnson et al., 2010). Raynor (2001) found significantly lower isometric and isokinetic force production in knee flexors and extensors of children with Developmental Coordination Disorder (DCD), a condition characterized by impairment in motor coordination. Weakness may contribute to decreased movement experience and low motor proficiency experienced by both children with DCD and NF1.

Hand held dynamometry is used clinically to evaluate muscular force because it is portable, lower in price than isokinetic equipment, and can be performed fairly rapidly. The inter-rater and intra-rater reliability of hand held dynamometry has been shown to be good to high when a standardized protocol is used (Macfarlane, Larson, & Stiller, 2008; Rose, Burns, Ryan, Ouvrier, & North, 2008) although isometric force production may not reflect functional strength. It is difficult to account for variability in the strength of different examiners; therefore we chose to collect our own control values. We also chose to evaluate the force production generated on dynamometry with force generated on a force plate to determine if there was a relationship between strength measured with a hand held dynamometer and strength measured in a functional activity. We found that there was a strong relationship between peak vertical ground reaction forces at take off in both hopping and jumping and the strength of knee extensors in the NF1 group. There was not a significant relationship between peak vertical ground reaction forces during hopping and jumping and hip extensor and ankle plantar flexor strength. These results suggest that lower extremity isometric strength is related to functional strength when muscles specific to the functional task are tested, i.e., forces at take off during jumping and hopping reflected only knee extensor strength.

There was not a statistically significant difference between peak vertical ground reaction force for hopping or for jumping. Additionally, there was not a statistically significant difference between hopping on the dominant and non-dominant legs. These results are similar to those found in another study where healthy adults performed equally well on dominant and non-dominant limbs during a hopping task on a force plate (McElveen, Riemann, & Davies, 2010). We had two children in the youngest age range who were unable to hop well; therefore we suggest using jumping as a functional task for assessing knee extensor strength since it is easier for young children to perform. However, hopping may be a useful task for assessing functional muscle capability depending on the research question asked and the impairments of the participants. Since our results demonstrated weakness in hip extensors, the addition of a hip extensor related task would have been valuable. The use of torque measurements may be an alternative method for adjusting HHD data instead of using a regression equation, although this method would not account for differences in sex

and weight. Another limitation was the lack of force plate data for jumping and hopping from a control group. A final limitation was that we allowed children to use their arms during jumping and hopping. The momentum from arm swing may have influenced the peak ground reaction forces generated at take off and the variance attributed to knee extensor force at take off may be a low estimate.

5. Conclusions

Children with NF1 had significantly lower force production with hand held dynamometry than controls for isometric hip extension, but force production for knee extensors and ankle plantar flexors was similar to those found in the control group. Additional research is necessary to determine if decreased muscular force in children with NF1 is a primary or secondary impairment, if strength declines with age, if there is a pattern of muscular weakness, and if weakness contributes to the common orthopedic impairments found in children with NF1.

We found a relationship between the force generated with hand held dynamometry and the ground reaction forces for hopping and jumping. Knee extensor strength was highly related to peak vertical ground reaction forces at take off during a vertical jump and hop. Peak ground reaction forces have been used in children to detect changes in athletic performance, however ground reaction forces have not been used to detect improvement in motor performance in children with neuromuscular conditions. Performing functional tasks such as jumping on force plates, may be a more sensitive method for quantifying strength and motor function given the presence of confounding factors when using hand held dynamometry. There was no difference between vertical ground reaction forces for jumping and hopping nor was there a difference between dominant and non-dominant limbs, suggesting that a jumping task, which was easier for our patients to perform, could be used along with dynamometry in quantifying the functional capacity of muscles and for documenting functional change.

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Research Highlights

- Provides further evidence of a strength deficit in individuals with NF1 by documenting proximal lower limb weakness.
- Demonstrates a relationship between body function/structure (knee extensor strength) and activity(jumping and hopping).
- Demonstrates a method to quantify gross motor performance in children with a neuromuscular health condition.

Fig. 1.

Time-force curves normalized by body weight for peak ground reaction force at take off of three participants representing good, average, and poor jump performance.

legend: 0 represents take off during jumping; * represents peak ground reaction force at take off

Good jump performance represented by a rapid increase to high peak force compared to a poor jump performance represented by a slow increase to a low peak force.

Table 1

Characteristics of participants with NF1 and controls used to compute strength values, means $\pm$ standard deviations

	N	Age (years + months)	Height (cm)	Weight (kg)	BMI (kg/m ²)
Females Control (SD)		10+1 $\pm$ 3.6	139 $\pm$ 16	33 $\pm$ 12	16.6 $\pm$ 2.9
Females NF1(SD)	24	8+4 $\pm$ 3.4	121 $\pm$ 16	23 $\pm$ 8	15.4 $\pm$ 1.7
Mean difference (SE)	13	1.8 (1.16)	18.6 cm (.01)*	10 (4.5)*	1.1 (.93)
Males Control	24	9+8 $\pm$ 3.10	139 $\pm$ 24	35 $\pm$ 18	17.1 $\pm$ 3.0
Males NF1	13	8+2 $\pm$ 2.5	129 $\pm$ 8	28 $\pm$ 12	16.8 $\pm$ 3
Mean difference (SE)		1.4 (1.16)	8.8 (6.6)	6.5 (4.5)	.31 (.94)
All (males and females)					
Control	48	9+11 $\pm$ 3.7	139 $\pm$ 21	34 $\pm$ 14	16.9 $\pm$ 2.9
NF1	26	8+3 $\pm$ 2.9	125.4 $\pm$ 17*	25.9 $\pm$ 10	16.1 $\pm$ 2.9

Legend: SD – Standard Deviation; SE – Standard error;

* – statistically significant differences at the .05 level.

Table 2

Dynamometry Protocol: Joint motion, participant position, and dynamometer placement by therapist.

Dynamometry Protocol		
Joint Motion	Position	Placement of dynamometer
Hip extension	Prone, knee flexed 90° and thigh extended off surface; pelvis stabilized	Resistance given posterior mid-thigh
Knee extension at 90°	Sitting, knee flexed 90°; stabilize thigh against the table	Resistance given anteriorly 2 inches proximal to lateral malleoli
Plantar flexion	Supine, knee extended; thigh stabilized	Resistance given across metatarsal heads

Table 3

Comparison of force produced by hand held dynamometry (HHD) in the NF1 group and derived force* of the control group.

Movement	Mean Newton (SD)	95% CI of the difference	P value	Effect Size
Hip extension			0.001	.99
NF1 group	81.35 N (36.37)	48.54 to 13.58		
Control group	112.38 N (25.31)			
Knee extension			0.67	.18
NF1 group	104.02 N (38.53)	25.33 to 16.52		
Control group	108.42 N (36.52)			
Ankle plantar flexion			0.52	.18
NF1 group	153.19 N (60)	35.81 to 18.43		
Control group	161.88 N (32.77)			

Legend: SD – Standard Deviation; N – Newton; CI – Confidence Interval

* Derived forces were determined by using the method described by MacWilliams et al. (2010) to match control subjects by sex, height, and weight.

Table 4

Comparison of mean peak vertical ground reaction forces normalized by body weight at take off during jumping and hopping for the NF1 group.

Task	Mean peak vertical ground reaction force at take off and SD	95% CI	P value	Effect size
Jump	630.5 N/kg (260)	-44.1 to 232.5	0.17	.39
Hop (limb with the greatest GRF)	535.3 N/kg (225.9)			
Hop (limb with the greatest GRF)	535.3 N/kg (225.9)	-99.9 to 154.9	0.67	.12
Hop (opposite limb)	507.8 N/kg (216.91)			

Legend: GRF – ground reaction force; SD – standard deviation; N/kg – Newton/kilogram.

Table 5

Relationship of mean peak vertical ground reaction force at take off to hand held dynamometry force of different motions in the NF1 group.

Functional Task	Hip Extension force	Knee Extension force	Ankle Plantar flexion force
Jumping GRF	$r^2 = .02$ ($p = 0.28$)	$r^2 = .49$ ($p < 0.01$)	$r^2 = .01$ ($p = 0.56$)
Hopping (Greatest GRF of 2 limbs)	$r^2 = .01$ ($p = 0.40$)	$r^2 = .71$ ($p < 0.01$)	$r^2 = .02$ ($p = 0.42$)

Legend: GRF – ground reaction force.