Energy consumption does not change after selective dorsal rhizotomy in children with spastic cerebral palsy

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AIM To determine whether energy consumption changes after selective dorsal rhizotomy (SDR) among children with cerebral palsy (CP).

METHOD We retrospectively evaluated net nondimensional energy consumption during walking among 101 children with bilateral spastic CP who underwent SDR (59 males, 42 females; median age [5th centile, 95th centile]: 5y 8mo [4y 2mo, 9y 6mo]) compared to a control group of children with CP who did not undergo SDR. The control group was matched by baseline age, spasticity, and energy consumption (56 males, 45 females; median age [5th centile, 95th centile]: 5y 8mo [4y 1mo, 9y 6mo]). Outcomes were compared at baseline and follow-up (SDR: mean [SD] 1y 7mo [6mo], control: 1y 8mo [8mo]).

RESULTS The SDR group had significantly greater decreases in spasticity compared to matched controls (−42% SDR vs −20% control, p<0.001). While both groups had a modest reduction in energy consumption between visits (−12% SDR, −7% control), there was no difference in change in energy consumption (p=0.11) or walking speed (p=0.56) between groups.

INTERPRETATION The SDR group did not exhibit greater reductions in energy consumption compared to controls. The SDR group had significantly greater spasticity reduction, suggesting that spasticity had minimal impact on energy consumption during walking in CP. These results support prior findings that spasticity and energy consumption decrease with age in CP. Identifying matched control groups is critical for outcomes research involving children with CP to account for developmental changes.

Cerebral palsy (CP) is a neuromuscular disorder caused by a brain injury at or near the time of birth, and is the most common pediatric disability in the US, affecting over 2 per 1000 live births.1,2 Fatigue is one of the top complaints of children with CP and their families.3 A likely contributor to fatigue is the amount of energy children with CP consume during daily activities, such as walking. There are many different metrics used to evaluate energy during walking. Clinically, the volume of oxygen consumed per unit time, often called ‘energy consumption’, is a widely used indicator of exertion.4 The energy consumption during walking for children with CP has been estimated to be two to three times that of typically developing peers.5–9 The cause of increased energy consumption is unclear.

Spasticity, defined as a velocity-dependent resistance to stretch,10 has been theorized as a cause of the observed increase in energy consumption in CP. Spasticity can cause an increase in overall muscle activity, which is thought to directly contribute to elevated energy consumption. Spasticity is observed in up to 80% of children with CP1 and is also common in other neurological disorders such as multiple sclerosis and spinal cord injury. Prior research has found that individuals with multiple sclerosis and spasticity also have energy costs two times that of typically developing peers, suggesting that spasticity may be an important determinant of high energy during walking.11

Selective dorsal rhizotomy (SDR) is a neurosurgical procedure where afferent nerve fibers in dorsal rootlets are cut in order to reduce efferent excitations.12 SDR has been shown to significantly reduce spasticity.13–15 Prior outcome studies have also suggested that SDR may reduce energy consumption.16,17 For example, Carraro et al. found that energy consumption was significantly reduced across multiple walking speeds after SDR for nine children with CP.16 While these studies seem to indicate that spasticity can be a contributor to elevated energy consumption in CP, there are several critical limitations to drawing causal conclusions from these prior studies. There are numerous other factors that could contribute to changes in energy consumption after surgery. For example, changes in walking patterns and speed18 after SDR could change joint moments and muscle demands.5 Energy consumption5 and spasticity19 are also known to decrease with age among children with CP. Evaluating energy consumption before...
and after procedures thus requires that covariate factors, such as speed and age, be considered. Identifying appropriate control groups of peers with CP provides one method to address these challenges.

The purpose of this study was to determine if spasticity is a significant contributor to elevated energy consumption among children with CP by investigating if reducing spasticity after SDR leads to lower energy consumption. We hypothesized that if spasticity contributes to energy consumption for children with CP, then an SDR should result in greater changes in energy consumption compared to matched controls with CP who did not undergo an SDR. Understanding the role of spasticity on energy consumption is important to inform treatments that aim to lower energy consumption, reduce fatigue, and increase quality of life for children with CP.

**METHOD**

Participants

We retrospectively identified individuals with bilateral spastic CP who underwent gait analysis at Gillette Children’s Specialty Healthcare (St. Paul, MN, USA) between 1994 and 2018. Inclusion criteria for this study were individuals younger than 18 years old who: (1) had a primary diagnosis of bilateral spastic CP; (2) underwent a bilateral SDR before the age of 12 years; (3) had at least one gait analysis before (baseline) and after (follow-up) SDR that included both energy consumption and Ashworth scores.

We also identified a control group of peers with CP that met the above inclusion criteria but did not undergo an SDR. This group was matched to the treatment cohort by baseline levels of age, energy consumption, and spasticity (Fig. 1). To identify matching peers between the control and SDR cohort we transformed all matching variables for each participant into a single summary score using an autoencoder.20 An autoencoder is a neural network that can be used for dimensionality reduction of complex data.21 For this research, the autoencoder was defined using age, energy consumption, and spasticity across all children with bilateral spastic CP who had previously received a gait analysis at Gillette Children’s Specialty Healthcare to define a summary metric of the variations in these dimensions across the population. The summary scores were then calculated for each child in the SDR group and a k-nearest-neighbors search algorithm was used to identify the closest matching peer with CP. If two children in the SDR group matched to the same nearest neighbor peer, we checked to determine if the second-nearest neighbor was an acceptable match. To be acceptable, the distance to the second-nearest neighbor had to be within the 95th centile of the distances between the SDR group and their first nearest neighbors.

This method was used to actively match participants based on age, walking speed, and spasticity. To avoid over-constraining the matching algorithm, we evaluated whether other important variables were passively matched. In this context, ‘passively matched’ means the baseline values of variables were matched between groups despite not being included in the active matching. Additionally, the resulting groups were constrained after active matching in order to limit the difference in follow-up time between matched pairs to less than 18 months. All matching variables were compared between groups to evaluate the similarity of the cohorts using Wilcoxon rank-sum tests.

**Spasticity**

To evaluate spasticity, we calculated a summary spasticity score for each individual. The score was derived from the Ashworth Scale of six right limb muscle groups using principle component analysis.22 The six muscle groups were hip adductors, hip flexors, hamstrings, rectus femoris, plantarflexors, and posterior tibialis. The Ashworth Scale is a discrete scale with five levels used to categorize spasticity.23 At Gillette Children’s Specialty Healthcare, the following Ashworth Scale definitions are used: (1) no increase in tone, (2) slight increase in tone, (3) more marked increase in tone, (4) considerable increase in tone, and (5) rigidity. This spasticity summary score is a weighted average and can be interpreted as an Ashworth score on a continuous 1 to 5 scale.

**Energy**

Energy during walking was assessed by converting the time rate of breath-by-breath oxygen consumption ($O_2^{gross}$) to energy consumption ($E^{gross}$) using the conversion rate of 21 Joules/ml.24 Both $O_2^{gross}$ and $O_2^{rest}$ were converted to gas volume expressed under standard conditions of temperature, pressure, and dry from testing conditions. The testing protocol consisted of a 6-minute over-ground barefoot walking trial preceded by a 3 to 10 minute rest period.4 Resting energy consumption ($E^{rest}$) was assessed during supine or sitting.4 We calculated net-nondimensionalized energy consumption as:

$$\text{Net - nondimensionalized energy consumption} = \frac{(E^{gross} - E^{rest}) \times \frac{1}{mg \sqrt{gL_{\text{leg}}}}}{mg \sqrt{gL_{\text{leg}}}}$$

where $m$ is body mass, $L_{\text{leg}}$ is the length of the leg, and $g$ is acceleration because of gravity.

**Strength**

Strength was also collected during the physical exam using a manual muscle test (MMT) for hip extensors, hip flexors, knee extensors, knee flexors, and plantarflexors. Like the method used for the summary spasticity score, an individual strength score was calculated for each individual for both the baseline and follow-up visits using principle component analysis. This MMT summary score is a weighted average and can be interpreted as an MMT score on a continuous 1 to 5 scale, where 1 is defined as ‘visible or
palpable contraction (no range of motion)’ and 5 is defined as ‘full range of motion against gravity’.

Walking speed
Walking speed was measured during the 6-minute over-ground barefoot walking trial. We computed nondimensionalized walking speed as:

$$\text{Nondimensionalized speed} = \text{speed} \times \left( \frac{1}{\sqrt{g \cdot \text{leg}}} \right)$$

Statistical analysis
We used Wilcoxon signed-rank tests ($z<0.05$) to compare changes in net-nondimensionalized energy consumption, spasticity, and walking speed between baseline and follow-up within each group (SDR and control). We then used Wilcoxon rank-sum tests ($z<0.05$) to compare change in net-nondimensionalized energy consumption, spasticity, and walking speed between the groups (SDR vs control). All values are reported as a median (5th centile, 95th centile), unless otherwise noted. All analyses were done using Matlab (MathWorks Inc., Natick, MA, USA).

RESULTS
Baseline comparison of groups
We identified 101 individuals with CP (59 males, 42 females) who met the inclusion criteria for the SDR group (age 5y 8mo [4y 2mo, 9y 4mo]; height 107cm [96.5cm, 133.2cm]; weight 17.7kg [14.1kg, 27.3kg]). Our control group consisted of 101 individuals with CP (56 males, 45 females) who did not undergo an SDR (age 5y 8mo [4y 1mo, 9y 6mo]; height 110cm [96.5cm, 131.3cm]; weight 18.8kg [13.8kg, 33.6kg]). The distributions for age, walking speed, net-nondimensionalized energy consumption, and spasticity at baseline were well matched between groups (Table 1, Fig. 1). There were no significant differences between groups for baseline net-nondimensionalized energy consumption ($p=0.6$) and baseline age ($p=0.8$, Table 2). There was a small difference between groups for baseline spasticity (SDR 2.2 [1.4, 3.3] and control 1.9 [1.2, 3.0]; $p<0.001$), with an effect size of 0.8. This difference in spasticity corresponds to less than half of an Ashworth level, below the threshold for clinical significance. The time between visits was similar between groups (SDR 1y 6mo [1y 1mo, 2y 8mo] and control 1y 6mo [0y 11mo, 2y 11mo]; $p=0.7$).

Spasticity
Children who underwent SDR exhibited greater reductions in spasticity at follow-up compared to matched peers with CP ($p<0.001$; Table 1 and Fig. 2). The baseline summary spasticity score was 2.2 (1.4, 3.3) for the SDR group and 1.9 (1.2, 3.0) for the control group. The SDR group exhibited a large decrease in spasticity after surgery, with a follow-up summary spasticity score of 1.2 (1.0, 1.7; $p<0.001$). The change in spasticity after SDR varied from -2.7 to 0 (-0.9
Our results demonstrate the critical importance of identifying and comparing to a cohort of peers with CP when looking at the impact of SDR on energy consumption. Further, we also evaluated changes in walking speed between visits. The baseline nondimensional walking speed was 0.32 (0.14, 0.42) and 0.30 (0.13, 0.42) for the SDR and control groups respectively. The follow-up nondimensional walking speed was 0.32 (0.12, 0.40) and 0.32 (0.13, 0.41) for the SDR and control groups respectively. There was no significant difference in walking speeds between groups at baseline (p=0.7) or follow-up (p=0.9). There was no significant change in walking speed for either group (SDR p=0.9, control p=0.5).

**DISCUSSION**

While SDR was effective at reducing spasticity, there was no associated decrease in energy consumption compared to matched peers with CP who did not undergo an SDR. We had hypothesized that if spasticity contributes to high energy consumption, then an SDR would result in lower post-treatment energy consumption compared to matched controls with CP who did not undergo an SDR. However, the change of energy consumption for the children with CP who underwent SDR was not significantly different from the change in energy consumption for the matched controls with CP. This indicates that spasticity is not a primary factor contributing to elevated walking energy in children with CP when compared to typically developing peers. The energy reduction at follow-up observed in both groups was consistent with the idea that energy consumption decreases with age among children with CP, independent of treatment.26

Prior studies have reported reductions in energy consumption after an SDR. However, these studies used either a typically developing control or no control group at all.13,15,16 For example, Trost et al. reported a 22% reduction in energetic cost after SDR.14 In their study, the average age (SD) before and after SDR was 2 years 3 months (2y 3mo) and 8 years 9 months (4y 0mo). This is an age span during which we would expect energy to decrease substantially among children with CP regardless of treatment. To our knowledge, no studies have included a control group of peers with CP when looking at the impact of SDR on energy consumption. Our results demonstrate the critical importance of identifying and comparing to a cohort of peers with CP when evaluating treatments to differentiate

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whether observed changes are due to the treatment or natural development.27

There are a few possible explanations for why spasticity does not affect energy consumption: (1) the additional muscle activity from spasticity does not increase energy consumption; (2) the additional muscle activity from spasticity is not large enough to have a large effect; (3) the spasticity measured by the Ashworth score does not reflect the muscle activity during gait; or (4) other factors beyond spasticity are the primary contributors to the elevated energy consumption in individuals with CP. Other possible contributors to elevated energy consumption in children with CP include poor selective motor control,28 excessive cocontraction for stabilization,29 and altered muscle properties.30 While SDR provides a platform to evaluate the impacts of spasticity, other strategies will be necessary to evaluate the relative importance of these other factors and to identify optimal strategies for reducing energy consumption for children with CP. This critical, as physical fatigue is prevalent and hinders participation and quality of life among individuals with CP throughout their lifespan.3,31

This study analyzed retrospective data from clinical gait analysis, which, like any retrospective study, has certain limitations. Energy consumption in these analyses is measured from a 6-minute walk test that does not control walking speed, mood, or other possible confounding factors. These analyses are conducted barefoot, which may not represent energy consumption during activities of daily living. Previous research has shown no significant difference in oxygen consumption between walking barefoot or with shoes.32 We matched our groups by baseline spasticity, age, and energy consumption. Walking speed was not actively matched, but we found no significant difference in speed between groups at baseline or follow-up. We limited the difference in time between visits for matched pairs to a maximum of 18 months. This resulted in a similar time between visits for both groups, and we found the results were insensitive to the selected threshold. We did not control for other surgeries that occurred between visits. Post hoc examination showed that 9% of the SDR group and 36% of the controls had orthopaedic surgery. Additionally, 7% of the SDR group and 29% of the controls received botulinum neurotoxin A (BoNT-A) injections. While BoNT-A may also impact spasticity, prior research has demonstrated that BoNT-A injections result in small and transient effects on spasticity. In a placebo controlled trial, BoNT-A provided minimal advantage compared to placebo at 4 and 6 weeks as measured by the Ashworth Scale (~0.25 reduction lower than placebo), and no advantage at 2, 4, 6, 8, or 12 weeks as measured by the Tardieu scale.33 A recent systematic review of long-term effects of SDR also showed the long-term effect of SDR on spasticity is unclear, with many studies reporting additional spasticity treatment.34 Results were similar between cohorts that either received or did not receive orthopaedic surgery or BoNT-A injections. There are concerns that SDR may cause weakness, and that this weakness may lead to increases in energy that could offset decreases from spasticity reduction. However, prior research has documented either no change or an increase in strength after SDR.35-38 We similarly found no significant changes in strength between visits for either group using values from MMT during the physical exam. MMT

Figure 2: Spasticity and net-nondimensionalized (NN) energy consumption for children with cerebral palsy (CP) who underwent a selective dorsal rhizotomy (SDR) and matched peers with CP who did not undergo SDR (control). (a) Baseline spasticity and NN energy consumption were similar between groups. Gray lines show normative values for typically developing (TD) peers from Gillette Children’s Specialty Healthcare. (b) Spasticity and NN energy consumption decreased significantly at follow-up for both groups. The SDR cohort had a significantly greater decrease in spasticity compared to the no-SDR group, but a similar decrease in NN energy consumption. Bars represent distributions for each group including outliers (*). [Colour figure can be viewed at wileyonlinelibrary.com]
provides limited precision for strength measurements, but is the most common strength measurement used in the clinic.

CONCLUSIONS

Spasticity was significantly reduced for individuals with CP who underwent an SDR compared to matched peers with CP, but energy consumption was not different between groups. These results demonstrate that spasticity has minimal impact on the high energy consumption observed among children with CP. Both groups demonstrated a reduction in spasticity and energy consumption between visits, likely reflecting changes due to development. Selecting appropriate control groups is critical for research involving children with CP to account for changes in function due to age, development, and other factors. While SDR is often suggested to reduce spasticity and improve energy, clinicians and families should understand that this procedure does not improve energy consumption during walking.

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