Total Energy Expenditure and Body Composition of Children with Developmental Disabilities

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Background
Obesity prevalence is increased in children with developmental disabilities, specifically in children with spina bifida and Down syndrome. Energy expenditure, a critical aspect of weight management, has been extensively studied in the typically developing population, but not adequately studied in children with developmental disabilities.

Objective
Determine energy expenditure, fat-free mass and body fat percentile and the impact of these findings on recommended caloric intake in children with spina bifida and Down syndrome.

Methods/Measures
This pilot study included 36 children, 18 with spina bifida, 9 with Down syndrome and 9 typically developing children. Half of the children with spina bifida were non-ambulatory. Doubly labeled water was used to measure energy expenditure and body composition. Descriptive statistics described the sample and MANOVA and ANOVA methods were used to evaluate differences between groups.

Results
Energy expenditure was significantly less for children with spina bifida who primarily used a wheelchair (p = .001) and children with Down syndrome (p = .041) when compared to children without a disability when adjusted for fat-free mass. However, no significant difference was detected in children with spina bifida who ambulated without assistance (p = .072).

Conclusions
Children with spina bifida and Down syndrome have a significantly decreased energy expenditure which directly impacts recommended caloric intake. No significant difference was detected for children with spina bifida who ambulated, although the small sample size of this pilot study may have limited these findings. Validating these results in a larger study is integral to supporting successful weight management of these children.

Keywords
Developmental disabilities, Spina bifida, Down syndrome, Obesity, Energy expenditure

The prevalence of overweight or obesity in children with the developmental disabilities (DD) of spina bifida (SB) and Down syndrome (DS) has been reported as two to four times higher than their typically developing (TD) peers. Large national surveys are not available for these populations. Reported estimates of overweight and obesity prevalence are based on convenience samples and may vary based on the measurement methodology performed. Currently, Body Mass Index (BMI) using standing height is commonly employed in clinical environments due to ease of use and cost-effectiveness. However, estimates of overweight or obesity based on
BMI in children with certain disabilities are inaccurate. It has been suggested that BMI criteria for obesity be modified for those with SB, but population-based studies are needed to confirm this approach. One study in youth with DS suggests that BMI accurately identifies excess body fatness in youth ≥95th percentile, but less accurately in those between the 85th and 95th percentile.

Excess body weight in children with DD creates the same risk for obesity-related co-morbidities such as cardiovascular disorders and type 2 diabetes as in TD children. Additional obesity-related concerns include decreased mobility and independence with self-care and increased risk of skin breakdown, social isolation, surgical complications and barriers to caregiver's providing daily care. A primary goal for children with DD is to facilitate independence and autonomy as they transition into adulthood. Related risks of obesity create unnecessary barriers to transition and can lead to negative self-management behaviors and poor outcomes.

The basis for the increased prevalence of obesity in children with DS and SB is multifaceted. As in the general population, understanding the balance of energy in (nutrition) and energy out (physical activity and basal metabolism) in children with DS and SB is essential. If energy intake is higher than energy expenditure, weight gain ensues. Confounding these primary issues are multiple factors inherent to the child's underlying condition that diminishes energy expenditure. Children with SB have: a) decreased lean body mass and basal metabolic rate, b) altered or reduced mobility dependent on the level of spinal lesion, and c) overall decreased energy needs, due to lower extremity paralysis, when compared with their TD peers. Children with DS often have reduced levels of physical activity, a lower basal metabolic rate secondary to less lean mass, increased incidence of hypothyroidism, and abnormal response to certain hormones (i.e. prepubertal children with DS have increased leptin that should increase the individual's satiety). These factors contribute to a lower energy expenditure which results in a reduced caloric need, increasing the risk for excess caloric intake and subsequent weight gain. In addition, parents and caregivers often overestimate the amount of energy expended through physical activity contributing to overestimation of energy needs. Although not evaluated in parents of children with DD, it is feasible to consider that this may occur in this population also.

Currently, total energy expenditure has been extensively studied in TD children but few studies have been reported in children with SB or DS. The knowledge of the child's energy expenditure, and subsequently recommended caloric intake, is an integral component of weight-related research, intervention development and provision of clinical care. The four groups in this study are children with: 1) SB who use wheelchairs (SB-WC), 2) SB who ambulate independently (SB-AMB) 3) DS (DS), or 4) no chronic illness (CON). The aims of this pilot feasibility study were to 1) compare total energy expenditure (TEE), body fat percentiles (BF%) and fat-free mass (FFM) by group, and 2) based on FFM, examine the impact of TEE on the child's energy needs in a sample of 36 children 4–18 years of age.

Methods
This study was part of a larger feasibility study that examined the use of various measures to assess energy expenditure in children with SB and DS. Internal Review Board approval was obtained through a Midwestern Children's Hospital.

Design/Setting
This cross-sectional pilot study was conducted within the Pediatric Translational Research Unit, a partner of the National Institutes of Health funded Clinical and Translational Science Institute of Southeast Wisconsin located within a Midwest Children's Hospital.
Participants
Study participants were recruited through mailings to patients of a local clinic, study advertisements posted in the clinic, social media sites, or SB and DS newsletters. Inclusion criteria were children with a diagnosis of SB, DS or no chronic illness; between 4 and 18 years of age and English speaking. Children who lived or were traveling >200 miles during the test period were excluded as differences in water across geographic regions could potentially impact doubly labeled water (DLW) analysis.28 The sample was stratified by group, age, and mobility status (Table 1). Prior to participation, consent and assent were obtained.

Table 1. Recruitment sample.

<table>
<thead>
<tr>
<th>Age of Child</th>
<th>Spina Bifida who Primarily use Wheelchair</th>
<th>Spina Bifida who Ambulate Without Assistance</th>
<th>Down Syndrome</th>
<th>No Chronic Illness</th>
</tr>
</thead>
<tbody>
<tr>
<td>4-7 years of age</td>
<td>3</td>
<td>3</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>8-12 years of age</td>
<td>3</td>
<td>3</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>13-18 years of age</td>
<td>3</td>
<td>3</td>
<td>3</td>
<td>3</td>
</tr>
</tbody>
</table>

Measures

Demographic questionnaire
Child's diagnosis, gender, race, ethnicity, age, method of mobility, family income, and marital status were collected from parent report.

Anthropometric assessments
Arm span was measured on all participants and a standing height (cm) was obtained if the child could stand independently. Arm span was obtained by two clinicians using a tape measure with the child sitting in a chair holding arms perpendicular to the floor. The measure was taken across the child’s back from the tip of the longest finger to the tip of the opposite longest finger. For height, participants stood erect with their back against the calibrated wall-mounted stadiometer and the headpiece lowered to the crown of their head. Each measure was performed three times and the average used. Body weight (kg) was obtained either by 1) a calibrated digital scale for children who could stand or 2) a wheelchair scale for those who used a wheelchair. In the latter, the child was weighed sitting in the wheelchair which was subsequently weighed alone and subtracted from the original combined participant and wheelchair weight.

Body Mass Index
The child’s height(s) and weight were used in the BMI calculation (kg/m²). The BMI was plotted on age and gender appropriate Centers for Disease Control (CDC) graphs to determine the BMI percentile and weight status classification. For children able to stand, BMI based on standing height and arm span were calculated. For children unable to stand independently, only arm span was used. Overweight status was identified as BMI ≥ the 85th percentile on the CDC growth charts.29

Total energy expenditure, body fat% and fat-free mass
Total energy expenditure is the amount of calories an individual expends daily and is a combination of an individual’s basal metabolic rate, energy spent on the thermic effect of food, energy expended during physical activity and for children the energy cost for growth. If calories are consumed above the TEE, weight gain is expected to occur.

Body composition, based on the two compartment model, is a combination of an individual’s body fat and FFM (water, bone, organs, and muscle). Body fat% and FFM were calculated based on total body water measures from DLW. At this time, there are no accepted cut-points for defining excess weight by BF% for children. For this
study, BF% cut-points proposed by Williams et al., (25% for boys and 30% for girls) based on cardiovascular risks and used with the Fitnessgram were used.\textsuperscript{30,31} We did not distinguish between overweight and obesity, but categorized weight as overweight vs. not overweight based on these cut-points.

Doubly labeled water is a gold-standard measure of TEE in free-living subjects. This measure adds two stable isotopes (deuterium and oxygen-18) to water and once ingested acts as a tracer. When combined with a change in body energy stores and measured (through blood, urine or saliva) over a period of time, it provides an accurate measure of TEE and total body water which can be used in calculations to determine fat mass and FFM.\textsuperscript{32}

For this study, participants fasted for 10–12 h prior to their morning appointment. The flavorless DLW was dosed per the individual's weight. Prior to the DLW ingestion, a urine sample was collected. Post DLW ingestion, urine samples were collected hourly for four voids prior to the child being sent home and twice by the family on days 7 and 14. Instructions and urine collection and mailing supplies were provided to the family along with phone calls from the study team. Samples were refrigerated and analyzed by isotope ratio mass spectrometry at the IRMS Core Laboratory of the Biotechnology Center in Madison, Wisconsin.

**Analysis**

Statistical analysis was performed with SPSS 22 (IBM SPSS Inc, Chicago, IL, USA). Descriptive statistics were used for demographic information and anthropometric measures of the 36 children (Table 2). For measures dependent on DLW (TEE, FFM and BF%), a cohort of 33 children was used due to quality control issues with three DLW samples. Of the 36 DLW samples, three were dropped from analysis because one urine sample was missing, one mislabeled and one compromised by contamination. While participants were stratified by age for recruitment, analysis was not conducted by age groups due to small sample size.

**Table 2. Participant characteristics.**

<table>
<thead>
<tr>
<th>Child Characteristics</th>
<th>Spina Bifida – Wheelchair M (SD)</th>
<th>Spina Bifida – Ambulatory M (SD)</th>
<th>Down Syndrome M (SD)</th>
<th>Control M (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Anthropometric Sample</td>
<td>n = 9</td>
<td>n = 9</td>
<td>n = 9</td>
<td>n = 9</td>
</tr>
<tr>
<td>Age (years)</td>
<td>11.22 (5.33)</td>
<td>10.33 (4.33)</td>
<td>10.00 (3.91)</td>
<td>11.22 (3.63)</td>
</tr>
<tr>
<td>Body Weight (kg)</td>
<td>36.91 (19.23)</td>
<td>40.3 (17.68)</td>
<td>34.32 (16.47)</td>
<td>43.59 (19.86)</td>
</tr>
<tr>
<td>Height (cm)</td>
<td>Stadiometer: 139.00 (24.96)</td>
<td>129.07 (24.44)</td>
<td>122.54 (24.10)</td>
<td>149.79 (24.61)</td>
</tr>
<tr>
<td>Arm Span</td>
<td>137.04 (27.41)</td>
<td>141.19 (28.27)</td>
<td>122.54 (24.10)</td>
<td>149.79 (24.61)</td>
</tr>
<tr>
<td>BMI%/Weight Category</td>
<td>Standing BMI%: 68.67 (26.75)</td>
<td>70.56 (23.78)</td>
<td>53.3 (27.14)</td>
<td></td>
</tr>
<tr>
<td>Not Overweight (&lt;85%)</td>
<td>5 (55.6%)</td>
<td>6 (66.7%)</td>
<td>9 (100%)</td>
<td></td>
</tr>
<tr>
<td>Overweight (≥85%)</td>
<td>4 (44.4%)</td>
<td>3 (33.3%)</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Arm Span BMI%</td>
<td>47.78 (44.5)</td>
<td>62.89 (32.33)</td>
<td>86.89 (9.39)</td>
<td>57.56 (29.84)</td>
</tr>
<tr>
<td>Not Overweight (&lt;85%)</td>
<td>6 (66.2%)</td>
<td>6 (66.7%)</td>
<td>4 (44.4%)</td>
<td>9 (100%)</td>
</tr>
<tr>
<td>Overweight (≥85%)</td>
<td>3 (33.3%)</td>
<td>3 (33.3%)</td>
<td>5 (55.6%)</td>
<td>0</td>
</tr>
<tr>
<td>DLW samples</td>
<td>n = 8</td>
<td>n = 7</td>
<td>n = 9</td>
<td>n = 9</td>
</tr>
<tr>
<td>Body Fat%</td>
<td>35.28 (15.17)</td>
<td>31.61 (8.94)</td>
<td>25.42 (10.03)</td>
<td>23.30 (5.96)</td>
</tr>
</tbody>
</table>
Not Overweight 0 2 (28.6%) 7 (77.8%) 7 (77.8%)
Overweight 8 (100%) 5 (71.4%) 2 (22.2%) 2 (22.2%)
Fat-Free Mass (kg) 20.75 (10.06) 26.84 (13.06) 25.47 (12.71) 33.91 (16.73)
Total Energy Expenditure (kcal/day) 1314.00 (448.89) 1836.14 (523.80) 1813.11 (549.08) 2646.33 (867.03)

BMI: Body Mass Index; DLW: Doubly Labeled Water; BMI% based on age and gender; Body Fat% cut-points of 25% for boys and 30% for girls.

For Aim 1, a one-way between groups multivariate analysis of variance (MANOVA) was executed to compare TEE, BF% and FFM among the four groups: SB-WC (n = 8), SB-AMB (n = 7), DS (n = 9) and CON (n = 9). Due to small sample size, we did not control for potential confounding variables (i.e. pubertal development, gender or age). No violations of normality, homogeneity of variance or multicollinearity occurred. A Bonferroni correction was used with the MANOVA due to three dependent variables evaluated with our small sample (significance level = 0.05/3 = 0.017). A one-way analysis of variance (ANOVA) was conducted with a post-hoc comparison (Tukey HSD) if the MANOVA was significant to determine which group differences were statistically significant (p ≤ .05). For aim 2, the TEE and FFM were plotted yielding a trend line for each group. The difference in TEE by group was calculated by subtracting the group’s TEE trend line from the control group matched on FFM.

Results

The cohort (N = 36) ranged in age from 4–18 years of age (10.7 yrs; [SD 4.1]; SB-WC; 11.2 yrs [SD 5.3], SB-AMB; 10.3 yrs [SD 4.3], DS; 10.0 yrs [SD 3.9] and Control: 11.2 yrs [SD 3.6]) and included 21 males (58%). The majority of participants reported their race as white (81%) followed by African American (8%), Asian (6%) and “other/did not answer” (6%). Two (6%) participants were of Hispanic/Latino ethnicity. Combined family annual income was high with 42% of the sample at $75,001-$100,000 and 33% between $30,000 and $75,000. The smallest percent were at the highest (> $100,000; 17%), and lowest (< $30,000; 8%) family incomes. Parents were mostly married (83%) and mothers primarily accompanied the child (97%). Anthropometric characteristics are presented including the percent of participants considered overweight based on the BMI% cut-point of 85% and BF% cut-points of 25% for boys and 30% for girls (Table 2). Due to small sample the equivalence of these measures were not evaluated.

Aim 1: Identify and compare TEE, FFM and BF% by group.

Participant's TEE, FFM and BF% were summarized by group (Table 2). The MANOVA analysis, evaluating these three dependent variables indicated a significant difference between groups (F (9, 66) = 4.90, p = .000; Wilks' Lambda = 0.29; partial eta squared = 0.34). However, only TEE was significant (F (3, 29) = 6.60, p = .002, partial eta squared = 0.41). A comparison of the mean group scores of TEE and FFM indicated that the Control group had the highest TEE and FFM followed by SB-AMB, then DS and finally SB-WC (Table 2).

The ANOVA for differences by group on TEE was significant F (3, 29) = 6.60, p = .002. Post-hoc comparisons indicated the TEE of the SB-WC (M = 1314; SD 449; 95% CI 938, 1689; p = .001) and DS (M = 1813; SD 549; 95% CI 1391, 2235; p = .041) groups were significantly lower than the control group (M = 2646; SD 867). However, no significance difference was detected between the control (M = 2646; SD 867) and SB-AMB group (M = 1836; SD 524; 95% CI 1352, 2321; p = .072).

Aim 2: Based on FFM, examine the impact of TEE on the child’s nutritional needs.

When mathematically matched for FFM, the recommended daily caloric intake, based on the difference between TEE averaged between 500 and 800 calories less per day for the DS and SB-WC groups compared to the control group (Fig. 1).
Discussion

There were a number of important findings identified in this pilot. Because growth and energy expenditure vary by ages, our ability to recruit children with DD across a wide age-range was important for future studies. Further, a difference in TEE for children from the SB-WC and DS groups was demonstrated. While not surprising that children with DS and SB-WC have a lower TEE, understanding how much less is imperative for successful weight management. No statistical significance was detected for the SB-AMB group, the small sample size may have limited the ability to detect a clinically significant difference. In fact, a sample of 108 would have been needed to determine a medium effect size with 80% power and the correction for multiple analyses. The results of this study will be useful in determining effect size for future work. In subsequent studies, with adequate sample size, analysis should include comparison of TEE by age groups in addition to diagnosis and mobility.

This difference in TEE between the groups may have major implications for nutritional counseling. The findings suggest that children with SB who use wheelchairs and those with DS may need 500–800 fewer calories per day than TD children to maintain their weight. While the sample is too small to draw absolute conclusions, a 500 calorie per day deficit in TEE over time without modifying energy intake can result in substantial weight gain. Without this awareness or early anticipatory guidance, inadvertent overfeeding is likely to occur further exacerbating weight issues. Future studies are needed to verify the decreased TEE and support clinicians and families to adopt management strategies in their daily lives. If findings are confirmed, incorporating a registered dietitian who specializes in counseling children with SB and DS and their families early on would be important for education on maximizing nutrition with a decreased caloric intake.

Weight status was categorized based on BF% (measured by total body water) and BMI. Although not tested due to sample size, the patterns of weight classification between these methods appear divergent regardless of how height was measured. The potential inaccuracy of BMI when classifying body composition in children with SB and DS has been questioned and further evaluation of this commonly used measure should be conducted.34,33

Strengths of the study include documenting the feasibility of using DLW, a gold-standard criterion measure, to examine energy expenditure in a population at high-risk for obesity. Additional strengths include the ability to recruit a sample stratified by age, diagnosis and mobility; essential for a larger population-based study. Limitations include a small sample. In addition, missing DLW data on three participants may have influenced the findings.
Conclusion
The findings from this pilot study highlight important obesity-related measurement issues in children with DD. Decreased TEE and subsequently reduced energy needs were identified. In addition, there is a lack of population-based studies and a need to identify a method to accurately identify weight status in a reliable and cost-effective manner. If confirmed in larger samples, the preliminary findings will impact future research, intervention development and clinical care.

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Disclosures
All authors have no disclosures or conflicts of interest to report.

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