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Body Composition Measurement in Children with Cerebral Palsy, Spina Bifida and Spinal Cord Injury: A Systematic Review of the Literature

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Abstract

Pediatric obesity is a major health concern that has an increased prevalence in children with special needs. In order to categorize a child's weight, an assessment of body composition is needed. Obtaining an accurate body composition measurement in children with special needs has many challenges associated with it. This perplexing scenario limits the provider's ability to screen, prevent and treat an abnormal weight status in this vulnerable population. This systematic review summarizes common methods of body composition measurements, their strengths and limitations and reviews the literature when measurements were used in children with cerebral palsy, spina bifida and spinal cord injury. Following PRISMA guidelines, 222 studies were identified. The application of the inclusion and exclusion criteria yielded a final sample of nine studies included in this review. Overall, articles reinforced the inconsistencies of body composition measurement and methodology when used with children with special needs. Concerns include small sample sizes, the need to validate prediction equations for this population, and the lack of controlled trials and reporting of measurement methodology. Healthcare providers need to be aware of the complexities associated with measuring body composition in children with special needs and advocate for further testing of these measurements. Additional studies addressing the reliability and validity of these measures are needed to facilitate appropriate health promotion in children.

Keywords

Adolescents; body composition; pediatric; special needs

The increased prevalence of pediatric obesity is recognized as a public health crisis that has the potential to lead to adverse morbidity and mortality in adulthood (Karnik & Kanekar, [20]). In 2011–2012, nationally 31.8% of children were either overweight or obese and 16.9% were classified as obese (Ogden, Carroll, Kit, & Flegal, [32]). Within the special needs (SN) population (individuals with physical and/or intellectual disabilities) the prevalence of obesity can vary by diagnosis, but in general it is higher than the general population (Yamaki, Rimmer, Lowry, & Vogel, [42]).

Obesity can be defined as having excess body fat or having body weight that is greater than what is appropriate for the individual's height (Centers for Disease Control, [7]). Body composition (BC) is a measurement of the proportion of fat, muscle, and bone and can be conveyed as body fat percentage and lean mass percentage (Toomey, Cremona, Hughes, & Norton, [39]). Body composition measurements are often used as a proxy for documentation of obesity (Rieken et al., [34]). Unfortunately identifying an accurate BC measurement in the SN population is complicated. The purpose of this review is to synthesize the evidence on BC measurements of children who are diagnosed with cerebral palsy, spina bifida, or spinal cord injury.

Background and Significance

Obesity has been described as a healthcare epidemic (Hurt, Kulisek, Buchanan, & McClave, [17]). In order to accurately classify an individual as overweight or obese, an accurate method for determining either fat mass compared to lean mass or body weight for height must occur. This is often done through BC measurement. The most accurate measure of BC is cadaver analysis which is not a viable option, so indirect measures of BC are used (Toomey et al., [39]). Indirect measures of BC are based on properties of fat free mass in the typically developing individual (Toomey et al., [39]) allowing a prediction of BC (Bila, 2013). Dependent on the diagnosis,

children with SN may have an altered body habitus that can include atypical fat and/or water distribution, short stature, contractures, kyphosis, scoliosis, and decreased muscle mass (Gurka et al., [15]; Kuperminc et al., [22]; Littlewood, Trocki, & Cleghorn, [25]; Liu, Roberts, Moyer-Mileur, & Samson-Fang, [26]). Associated orthopedic complications or mobility limitations can lead to additional challenges in obtaining an accurate height (Hogan, [16]; Veugelers, Penning, van Gulik, Tibboel, & Evenhuis, [40]). Since the measurement of height is an integral component of many BC measures, any inaccuracy in this measure will affect the accuracy of the BC. This concern has led to the use of proxy measurements of height such as the measurement of arm span, segmental length, and recumbent length (Froehlich-Grobe, Nary, Van Sciver, Lee, & Little, [13]). Each of these proxy measures has its own limitations.

Prediction equations are integrated within BC measures to determine body fat percentage. Prediction equations work well for the population they were derived from, usually healthy young or middle aged populations. Acknowledging this limitation, researchers often create new prediction equations based on the sample being studied (Casey, [<u>6</u>]). However, before being considered a reliable equation further testing and validation in the specific sample must occur which is often not completed. Currently, there is a lack of validated equations for use in children with SN.

Common methods of measuring BC include the use of a calculated body mass index, skinfold thickness, or bioelectrical impedance analysis. Common criterion measures include a dual-energy x-ray absorptiometry scan, and isotope dilution. These measures along with their strengths, limitations, and specific concerns when used in the SN population are delineated in Table 1.

Table 1. Summary of common body composition and criterion measures.

| Common Body Composition Measures | | | | |
|--|--|---|---|--|
| Measurement | How it Works | Strengths | Limitations | Special Needs Concerns |
| Body Mass Index (BMI) | Calculation of Kg/m ² = BMI. For individuals 2-20 years of age, a BMI is placed on a graph specific to their age and gender. BMI between 85% and 95% is overweight and ≥95% is obese. | Simple Cost effective Easy to use Non-invasive Clinically accessible Well accepted for individuals 2 years of age and older. | Does not differentiate between the individual's muscle and fat mass. Accuracy depends on height and weight being accurate. | Children with atypical body habitus can have increased regional adiposity, but still have an acceptable or low BMI. Obtaining an accurate height can be challenging secondary orthopedic abnormalities or the inability to stand independently. |
| Skinfold Thickness (SF) | Handheld calipers are used to measure subcutaneous fat at different anatomical sites and the measurements are put into a prediction equation to determine body density and then a second equation for body fat %. | Cost effective Non-invasive Clinically accessible | Prediction equation uses two calculations increasing risk of error. Increased risk of error based on technician's training, individual's hydration status, and skin thickness. Increased inaccuracy with higher body fat%. | Lack of population specific prediction equations for children with special needs. |
| Bioelectrical Impedance Analysis (BIA) | Estimates total body water and subsequently body fat % by measuring the impedance of electricity by body tissues. Electrical current flows more rapidly through fat free mass than adipose tissue and bone allowing total body water to be determined. Formula used requires height and weight of individual to be inputted into the analyzer. | Ability to be performed in standing or recumbent position Cost effective Non-invasive | Accuracy depends on individual's fluid distribution, specific testing methodology (fasting, body positioning, dehydration status, localized changes in extremities [edema] and recent exercise), accurate height and weight being used and body symmetry. | Fluid distribution can vary in certain populations such as spinal cord injury or spina bifida. Significant body asymmetry such as secondary amputation, unilateral hemiparesis and neurological |

| | Clinically accessible | Equations used are based on population specific testing. | muscular conditions can impact accuracy. |
|--|--------------------------|--|---|
| | | | Lack of population specific prediction equations for children with special needs. |

Table 1. Summary of common body composition and criterion measures.

| Common Body Composition Criterion Measures | | | | |
|--|--|---|---|--|
| Measurement | How it Works | Strengths | Limitations | Special Needs Concerns |
| Dual-Energy X-Ray Absorptiometry (DXA) | Participant lies in supine position on table and DXA scan uses a small dose of radiation to provide estimate of body fat, lean mass, bone mineral content and bone mineral density. | Considered a valid and reliable measure of body composition. Yields regional and whole body values. Can be used in all age groups. | Expensive Not clinically accessible Need trained operators Limitations with metal implants Use of radiation limits repeated use | Children with contractures may be uncomfortable and unable to lie still for measurement on table. |
| Isotope Dilution | Deuterium oxide or other stable isotopes are mixed in water and ingested and then measured through blood, saliva or urine as it is excreted from body. Allows the estimation of total body water and subsequently body fat%. | Considered a valid and reliable measure of body composition. | Expensive Not clinically accessible Need trained experts to supply the water and analyze the results | Children may not be able to take in oral liquids. Total dose needs to be ingested, which may be difficult for participants with swallowing or drooling. Can be used with a gastrostomy tube if dose is followed by a flush to ensure total dose is ingested. |

3 Alschuler, et al. ([1]); Ball, Swan, & Altena ([2]); Barreira, Renfrow, Tseh, & Kang ([3])' Bila, et al. ([5]); Clasey & Gater ([9]); de Menezes, Souza Lopes, Cunha, Jansen, & dos Santos ([10]); Gater ([14]); Gurka, et al. ([15]); Hogan ([16]); International Atomic Energy Agency (1999); International Atomic Energy Agency ([19]); Kispert & Merrifield ([21]); Kushner, Gudivaka, & Schoeller ([23]); Lee & Gallagher ([24]); Mialich, Sicchieri, & Jordao ([28]); Miyatani, Yang, Thomas, Craven, & Oh ([30]); Mojtahedi, Valentine, & Evans ([31]); Rombeau, et al. ([36]); Talbert, et al. ([38]); Veugelers, Penning, van Gulik, Tibboel, & Evenhuis ([40]); Wells & Fewtrell ([41]).

Special Needs Populations

Children with SN have an increased risk for obesity; however the accuracy of population level data is questioned (Rimmer, Wang, Yamaki, & Davis, [35]). Many national prevalence estimates are based on self-reported height and weight data, which are associated with higher error rates or due to challenges in measurement may be using an inaccurate height or weight (Rimmer et al., [35]). These concerns lead to statistics that may underrepresent obesity in these populations (Rimmer et al., [35]). Three exemplar conditions addressed in the literature are cerebral palsy (CP), spina bifida (SB) and spinal cord injury (SCI).

Cerebral palsy has associated motor impairments or loss of mobility, short stature, lower fitness, and less muscle mass leading to an increased risk for obesity (Gurka et al., [15]; Kuperminc et al., [22]; Peterson, Gordon, & Hurvitz, [33]). Kyphosis and scoliosis can occur in more severe CP which can hinder the ability to obtain an accurate height measurement (Veugelers et al., [40]). In addition, children with CP can have an altered energy metabolism, general nutritional problems such as poor appetite, dysphagia, constipation, and hypersensitivity of the oropharynx which can lead to malnutrition (Veugelers et al., [40]). Each of these complications can alter an individual's hydration status which can lead to error in most measures of BC.

Spina bifida is a birth defect that occurs when the neural tube closure is impaired during early pregnancy (Mita et al., [29]). Obesity prevalence in individuals with SB has been estimated as high as 64% among adolescents (Dosa, Foley, Eckrich, Woodall-Ruff, & Liptak, [11]). Associated deterioration of lower extremity muscles, short stature, sedentary lifestyle, less lean body mass, and a lower basal metabolic rate have all been implicated in the risk or cause of the abnormal weight status in the individual with SB (Dosa et al., [11]; Mita et al., [29]). In addition, children with SB have an atypical distribution of body water which impacts the child's hydration status and complicates the measurement of total body water and subsequently BC (Littlewood et al., [25]).

After a SCI, the body undergoes changes including an increase in adipose tissue which may be related to overfeeding during the acute phase of SCI and rehabilitation, marked sarcopenia associated with muscle paralysis or muscle atrophy, and a decrease in energy metabolism (Gater, [14]). Individuals with SCI have increased adipose tissue and less lean muscle mass when compared to their general population counterpart, even at a normal Body Mass Index (BMI) (McDonald, Abresch-Meyer, Nelson, & Widman, [27]).

Methods

Search Strategy

This review of literature focused on BC measurement in children with CP, SB, and SCI. The strategy employed for this review included searching the following databases: PubMed, CINAHL, Cochrane Library, Healthstar, EMBASE, Web of Science, SPORTdiscus, and Google Scholar. Search terms included obesity, overweight, weight status, body composition, fat mass, lean mass, muscle mass, neural tube defects, anthropometrics, bod pod, segmental length, recumbent length, stadiometer, body mass index, DXA scan, bioelectrical impedance analysis, air displacement plethysmography, skin folds, limb length differences, adipose, adiposity, body composition study, waist circumferences, lipid profile, and hydrodensitometry. Each of the terms was combined with the following conditions: cerebral palsy, spina bifida (myelomeningocele), and spinal cord injury. Terms were searched as keywords and, when available, were mapped to MeSH and CINAHL subject headings. The search was limited to literature published between 2003 and October of 2013. Studies were included if they used a measurement of BC with a criterion or gold standard measurement for comparison in children/adolescents (up to age 21 years) with the diagnosis of CP, SB, or SCI. Commentaries, literature reviews, and articles that primarily focused on adults were excluded. A review of the retrieved studies' reference lists was performed to determine if additional studies could be identified.

The search identified 222 articles and 10 additional articles were identified through examination of references. An additional study that examined the use of arm span measurement as a substitute for height and then evaluated how the CDC/NCHS BMI graph could appropriately be used in children with SB was included in this review. While this study did not use a criterion measure, it was seen as a seminal study. The final sample for this review was 9 articles (see Figure 1).





These studies were evaluated using the hierarchy of evidence described by Fineout-Overholt, Melnyk, Stillwell, and Williamson ([12]). Each article underwent an independent review by authors Meyer-Wentland and Polfuss. Discrepancies or disagreements were discussed with author Sawin for final determination. All included articles were case control or cohort studies and were classified a IV (Fineout-Overholt et al., [12]). The evidence table, divided by diagnoses, included studies investigating BC in children with CP (n = 5), SB (n = 2) and SCI (n = 2) (see Table 2).

| First author (year) and level of evidence | Study aims | Study design and level of evidence | Sample | Major variables and measurement | Data analysis | Findings | Benefits and limitations |
|--|--|---|---|---|--|---|--|
| Cerebral palsy | | | | | | | |
| Bell, 2013 IV | Determine: Criterion validity of three different equations (Kushner, Pencharz, and Fjeld) to estimate TBW from BIA. Optimal electrode placement in unilateral CP Reliability of BIA measurements | Descriptive design as part of longitudinal study | 55 preschool aged children with unilateral and bilateral impairment secondary to CP 38M/17F | Functional severity (GMFCS) Weight (g) chair scale or portable electronic scale TBW and BF%—BIA Deuterium-dilution technique—TBW Height or length (depending on ability to stand) (mm) portable stadiometer or length board or knee height | Paired t-test Bland and Altman plots Regression Analysis | The Fjeld equation was most accurate in estimating TBW, but individual results varied up to 18%. A population specific equation was developed based on study Electrode placement can be on either side of body to determine TBW in unilateral CP. Duplicate measures should be performed for reliability in routine practice to ensure accuracy | Limitation of small sample size and while Fjeld equation was accurate at the population level should be used with caution at individual level New equation (population specific) was developed but needs to be validated in subsequent study Demonstrates use of BIA as a useful and accurate tool for the assessment of body composition in preschool aged children with CP. |
| Rieken, 2011 IV | Validate CP specific equations for EE (Krick, et al.) against DLW. Validate CP for specific equation for SF (Gurka, et al.) against isotope dilution. If either are inaccurate develop new equations | Cross sectional descriptive study | 61 children aged 2–19 years old with severe neurologic impairment and ID 32 M/29 F | DLW—TEE Tibia length (cm) as proxy for height Weight (kg) wheelchair or sling scale Height—crown to heel with flexible tape measure and child in recumbent position | Independent t- test ICC Bland and Altman plots Regression analysis | TEE was overestimated by Krick et al equation and TBW was underestimated by using Pencharz et al. equation. %BF overestimated by Gurka et al. equation based on SF SF not feasible in all children | Created equation specifically for target population based on objective measurement of TEE—will need further testing and validation Validity and feasibility of use of SF measurements was found to be less accurate in children with |

Table 2. Studies addressing measurement of body composition in children with cerebral palsy, spina bifida, and spinal cord injury.

| | | | | SF (4 sites—triceps, biceps, subscapular and suprailiacal) TBW and BF%—BIA EE (Krick et al.; accelerometers) Mobility (GMFCS) | | BIA estimated BF% better than SF Two new prediction equations created. Predicting TBW from BIA and tibia length (proxy for height) provided statistically significant predictor of TBW. To predict TEE, new equation using TBW was accurate. | severe neurologic impairment and ID Use of tibia length may be an accurate proxy for height Small sample size |
|-----------------------|---|---|---|---|--|---|--|
| Kuperminc, 2010 IV | Compare single anthropometric measures to criterion %BF in DXA Compare findings to those in typically developing children | Cross sectional descriptive sub- study of the North American Growth in Cerebral Palsy Project | 58 children 8–18 years old with CP GMFCS levels III, IV, and V. 33M/25F μ age 13y 1 mo ± 3yrs Compared to 6754 typically developing children from 1999-2004 NHANES | Weight (seated scale) Skinfold (triceps) Height (knee height) BMI (weight ² /height) Arm circumference Mid-upper arm fat area (by formula) Functional motor impairment (GMFCS) Sexual maturation (Tanner stage) %BF (Whole body DXA - Hologic Delphi W machine) | Linear regression R ² values Agreement analysis with 3x3 contingency table (weighted kappa statistic) | Children with CP and controls had similar levels of %BF by DXA, but children with CP had overall lower BMI, mid upper arm circumference, triceps SF and mid-upper arm fat area z-scores than control group. BMI z-scores had a low to moderate correlation to BF All anthropometric measures for children with CP and control group showed poor agreement with %BF by DXA, but agreement was worse for children with CP who had excess BF according to DXA | Single anthropometric measures are poor predictors of %BF for children with CP Recommended use of Slaughter equations with correction factor (not tested in study) with two SF sites. Limitation of small sample and limited generalizability by only including children with GMFCS levels III to V Potential inaccurate height measurement due to orthopedic problems or poor cooperation |
| Gurka 2010 IV | Confirm that Slaughter equations underestimate %BF in children with CP | Cross sectional descriptive sub- study of the North American | 71 children 8–18 years old with CP GMFCS levels I–V | Functional motor impairment (GMFCS) | Bland and Altman Plots CCC | Slaughter equations overall underestimated %BF compared to the %BF from DXA | Benefits included use of ambulatory and non- ambulatory children with CP and the use of |

| | If findings were inaccurate, then employ correction to the Slaughter equations | Growth in Cerebral Palsy Project | 41M/30F | Sexual maturation (Tanner stage) SF (Mean of two measurements of subscapular and triceps skinfolds) %BF (Whole body DXA - Hologic Delphi W machine) | | Use of Slaughter equation with a correction factor resulted in much improved predictive ability | DXA as criterion measure for %BF Correction factor is a simple addition of second step to Slaughter equation, which is good but can be another potential for error. Limitations include new formula needs validation and may be less accurate for larger individuals Small sample limits generalizability. |
|-----------------------|--|---|--|---|------------------------------|---|--|
| Liu, 2005 IV | Determine if correlation of BIA and anthropometry when compared to DXA for determination of FFM, BF, and %BF is adequate for clinical and/or research purposes | Cross sectional descriptive study | 8 children μ age 10 years with CP μ GMFCS 4.6 6M/2F | Functional Motor Impairment (GMFCS) Severity of CP (developmental pediatrician) Height (knee height) %BF (SF measures from 4 sites. Performed 2 and 4 site Slaughter and Durnin equation) BIA—Whole body resistance and reactance, FFM, BF, %BF FFM, BF, %BF (DXA— Hologic 4500A machine) | Spearman rho coefficients | Correlation coefficients were excellent for FFM for all methods, moderate for FM and %BF BIA performed best for determining FM and SF equations were best for determining %BF No advantage in using 4 site SF measurements vs 2 site SF measurements | BIA highly correlated with DXA for FFM Small sample size Need further study Children fasted before BIA which authors questioned that it may have disrupted TBW along with possibility of dehydration affecting lean body mass and overestimating FM |
| Spina Bifida | | | | | | | |
| Shurtleff, 2010 IV | Determine the most appropriate clinical method for calculation | Retrospective chart review from 7/65–6/08 | Patients seen in the Washington | Height/linear length (stadiometer in standing position if able to stand. | Descriptive statistics | Comparison of BMI means of patients with MMC suggest that using | Provides alternative option of using BMI growth charts and |

| | of BMI to monitor for | | Birth Defects | If unable to stand, supine | Bland and | arm span as a proxy for | ability to use arm span |
|-------------|-----------------------|-------------------|-------------------|-----------------------------|-----------------|----------------------------|---------------------------|
| | obesity and/or a | | Clinic | stadiometer and/or | Altman analysis | height is acceptable for | in place of height for |
| | propensity toward | | | segmental | | the CDC/NCHS BMI | children who are least |
| | obesity in patients | | 4,968 visits from | measurements from hip | | graphs for children with | paralyzed |
| | with MMC | | 709 patients. | to knee and knee to sole | | lower level lesions. | . , |
| | | | | of heel added to sitting | | | Demonstrates that arm |
| | | | | height) | | For patients with MMC | span used alone is a |
| | | | | | | and higher level lesions | useful approximation of |
| | | | | Weight (kg) platform | | there can be falsely low | height for children with |
| | | | | scale for those able to | | BMIs when placed on | MMC and lower level |
| | | | | stand and an infant or | | CDC/NCHS BMI graph | lesions. Recommends |
| | | | | wheelchair scale for | | which is thought to be | additional modifications |
| | | | | those unable to stand | | related to lower | for children with higher |
| | | | | | | extremity deformities or | level lesions. |
| | | | | Arm span (cm) measured | | scoliosis. | |
| | | | | with a metal ruler | | | |
| | | | | | | Falsely low BMIs in higher | |
| | | | | BIVII (calculated using | | level lesions can be | |
| | | | | both height and arm | | corrected by using arm | |
| | | | | span) | | span and lowering BMI | |
| | | | | I OL (Level 1: thoracic and | | percentiles on graph by | |
| | | | | high lumbar motor levels | | 50%. | |
| | | | | 1_3. Level 2. lumbar | | | |
| | | | | motor levels 4–5 and | | | |
| | | | | Lovel 2: sacral motor | | | |
| | | | | function lovals) | | | |
| | | | | runction levels) | | | |
| Littlewood, | Determine the | Cross sectional | 14 children μ age | LOL High:T1-L2; Mid: L3- | Descriptive | Measured TBW by | Cornish equation |
| 2003 IV | accuracy of current | descriptive pilot | of 8.49 ±3.85 | L4; Low: L5-S2 | statistics | deuterium dilution had | worked well at |
| | predictive equations | study | years diagnosed | | | the most significant | predicting TBW |
| | based on BIA in | , | with repaired | Ambulation Sacral-L5: | Bland and | correlation to predicted | |
| | determining TBW as | | MMC | community ambulators; | Altman analysis | TBW using Cornish | TBW from BIA |
| | compared to criterion | | | L4: functional | Completions | Equation (an equation | measurement could be |
| | of deuterium dilution | | 4 subjects with | ambulators; L3 and | Correlations | based on adults) vs | useful in children with |
| | | | high lesion; 3 | higher: non-functional | | Davies equation (an | MMC and could be used |
| | | | subjects mid- | ambulators | | equation based on | in clinical setting |
| | | | lesions and 7 | | | children and adolescents) | |
| | | | subjects low | TBW (BIA machine) | | | Needs further study |
| | | | lesion | TBW (Deuterium dilution) | | | with increased sample |
| | | | | | | | size |
| | | | 7M/7F | | | | Limitations include small |
| | | | | | | | sample size and |
| | | | | | | | sample size and |

| Spinal Cord | | | | | | | decreased generalizability Deuterium alone only gives measure of TBW not ratio of ICW/ ECW |
|-----------------------|---|--------------------------------------|---|--|--|---|---|
| Mojtahedi, 2009 IV | Compare %BF estimates from SF and BIA measurements using prediction equations with DXA estimates in athletes with SCI | Cross sectional descriptive study | 16 Caucasian athletes with μ age of 22.0 ±2.7 years of age with SCI Time since injury with μ 16.2 ±5.7 years Injury level range T5-L5 8M/8F | Physical activity level (PASIPD) Weight (nearest 0.1kg on beam scale adapted for nonambulatory individuals) Height (Gulick II retractable measuring tape from top of head to bottom of heel while in supine position. If unable to extend joints, measurement were taken in segments) BMI (weight in kg/m ²) Circumferences (measured to nearest 0.1 cm at umbilicus and hip [iliac crest] while in supine position) SF: (9 sites—triceps, subscapular, biceps, chest, midaxillary, paraumbilical, suprailiac, thigh and lateral calf) %BF BIA and DXA [Hologic QDR 4500A machine]) | Descriptive statistics Independent sample t-test Shapiro-Wilk test statistics Paired t-tests Bivariate correlations Regression analysis performed on Bland and Altman plots | All field methods underpredicted the %BF when compared with DXA BIA %BF estimates were closer to measured DXA %BF than SF estimates but had high variability, increased total error and poor correlations Of the SF and BIA prediction equations (Jackson and Pollock 7 site, Jackson, Pollock, and Ward 3 site, Evans et al. 7 site, Evans et al. 3 site and Siri equation), Evans et al.'s 3 site SF provided the best fit for this population | Benefit: Small sample size may have contributed to high variability in %BF estimates Unable to obtain participants ASIA impairment classifications, limiting the description of the arrange of injury levels Decreased generalizability with all subjects being college aged athletes Unknown potential error caused by spinal rods in 9 of 16 subjects Needs further research in this population with larger sample to develop more appropriate prediction equations for BC assessment |

| Height (wall mounted | Analysis of | No differences of height, | Benefit of showing |
|--|---|--|--|
| stadiometer if able to | covariance | weight, BMI or BMI% | reduced lean tissue |
| stand erect; supine | adjusted for | adjusted for age between | mass and significantly |
| stadiometer if unable to stand erect) Weight (Wheelchair balance scale for participants in wheelchair and if able to stand) BMI (calculated body weight kg/m ²) %BF (DXA [Hologic QDR 4500A]) Obesity (% trunk fat determined by DXA with % trunk fat ≥ 30% in males and ≥35 % in females considered obese) | age and gender Tukey multiple comparison analysis Regression analysis ROC curve | retraplegia group had significantly lower BMI and BMI% adjusted for age than control group Paraplegia and tetraplegia group had less lean tissue mass and more FM than control group %BF was significantly higher than control group for all BMI groups (average of 38% ±4% more fat tissue per unit BMI in SCI group vs. control group ROC found optimal criterion points for obesity to be 25 kg/m ² and 19kg/m ² for SCI | higher FM for children with SCI vs control with offer of new cut off point for obesity by BMI Questions accuracy of DXA readings of fat and lean tissue composition in pixels containing bone Recommends further studies with larger sample sizes to be conducted using a 4 compartment model to determine appropriate measures of obesity |
| | Height (wall mounted stadiometer if able to stand erect; supine stadiometer if unable to stand erect) Weight (Wheelchair balance scale for participants in wheelchair and if able to stand) BMI (calculated body weight kg/m ²) %BF (DXA [Hologic QDR 4500A]) Obesity (% trunk fat determined by DXA with % trunk fat ≥ 30% in males and ≥35 % in females considered obese) | Height (wall mounted stadiometer if able to stand erect; supine stadiometer if unable to stand erect)Analysis of covariance adjusted for age and genderWeight (Wheelchair balance scale for participants in wheelchair and if able to stand)Tukey multiple comparison analysisBMI (calculated body weight kg/m²)Regression analysis%BF (DXA [Hologic QDR 4500A])ROC curveObesity (% trunk fat determined by DXA with % trunk fat \geq 30% in females considered obese)Analysis of covariance adjusted for age and gender | Height (wall mounted stadiometer if able to stand erect; supine stadiometer if unable to stand erect)Analysis of covariance adjusted for age and genderNo differences of height, weight, BMI or BMI% adjusted for age between groupsWeight (Wheelchair balance scale for participants in wheelchair and if able to stand)Tukey multiple comparison analysisTetraplegia group had significantly lower BMI and BMI% adjusted for age than control groupBMI (calculated body weight kg/m²)ROC curveParaplegia and tetraplegia group had less lean tissue mass and more FM than control groupObesity (% trunk fat determined by DXA with % trunk fat ≥ 30% in males and ≥35 % in females considered obese)%BF was significantly higher than control group for all BMI groups (average of 38% ±4% more fat tissue per unit BMI in SCI group vs. control groupROC found optimal criterion points for obesity to be 25 kg/m²and 19kg/m²for SCI |

4 Legend: %BF: Percentage Body Fat; ASIA: American Spinal Injury Association; BIA: Bioelectrical Impedance Analysis; BC: Body Composition; BF: Body Fat; BM: Body Mass; BMC: Bone Mineral Content; CDC/NCHS: Centers for Disease Control and Prevention and National Center for Health Statistics; cm: centimeter; CP: Cerebral Palsy; CCC: concordance correlation coefficient; DXA: DXA Scan; DLW: Double Labeled Water; EE: Energy Expenditure; EI: Energy Intake; ECW: Extracellular Water; FM: Fat Mass; FFM: Fat Free Mass; F; female; g; gram; GMFCS: Gross Motor Function Classification System; ID: Intellectual Disability; ICW: Intracellular water; ICC: Intraclass correlation coefficients; kg: kilogram; LOL: Level of lesion; M: males; MMC: Myelomeningocele; NCHS: National Center for Health Statistics; PASIPD: Physical Disabilities Activity Scale for Individuals with Physical Disabilities; REE: Resting Energy Expenditure; ROC Receiver Operating Characteristic; SB: spina bifida; SCI: Spinal Cord Injury; SF: Skinfold thickness; TBW: Total Body Water; TEE: Total Energy Expenditure; WHO: World Health Organization; Level IV evidence: Case controlled or cohort studies. Equations that were listed in the table should be referenced as: Cornish (as cited in Littlewood, 2003), Davies (as cited in Littlewood, 2003), Evans et al. (as cited in Mojtahedi, 2009), Gurka et al. (as cited in Rieken, 2011), Jackson and Pollock (as cited in Mojtahedi, 2009), Jackson, Pollock and Ward (as cited in Mojtahedi, 2009), Krick et al. (as cited in Rieken, 2011), Siri (as cited in Mojtahedi, 2009).

Results

The findings are presented in two sections. First, study protocol methodology is discussed followed by study design, sample, criterion measures, findings, and limitations within each diagnosis group of CP, SB, and SCI.

Study Methodology

All five of the studies that included BIA measurements used the whole body technique versus a handheld model, but used different procedures. Two of the studies had participants lie in the supine position for 10 minutes prior to testing (Mojtahedi, Valentine, & Evans, [31]; Rieken et al., [34]). In contrast, Littlewood et al. ([25]) conducted the BIA test as soon as the child was in the supine position to avoid fluid shifts and two studies did not report on the length of time in supine position before testing (Bell, Boyd, Walker, Stevenson, & Davies, [4]; Liu et al., [26]). There was high variability in reported fasting status. Three studies had participants fast, one for at least 4 hours (Rieken et al., [34]), the second for 12 hours (Mojtahedi et al., [31]), and the third for a "period of time" before the test (Liu et al., [26]). The remaining two did not report on fasting status (Bell et al., [4]; Littlewood et al., [25]). When identified, exclusion criteria included: abnormal hydration status (Rieken et al., [34]), and children who were acutely ill (Littlewood et al., [25]).

The measurement of SF had wide variation amongst the five studies that included its use. Number of sites measured ranged from 1 to 9. When converting the body density to body fat percentage, six different prediction equations were used including Slaughter, Slaughter-Durnin, Siri, Evans et al., Gurka, and a correlation of a single SF measurement. Three of the five studies designated what side of the body the measurements were taken: right (Mojtahedi et al., [31]), left (Rieken et al., [34]) or the unaffected side (Gurka et al., [15]).

The largest inconsistency in measurement among the studies was height. Within the CP studies there were five different measures of height used based on the child's ability to stand (see Table 2). One study did not document how the height measurement was obtained (Gurka et al., [15]). Methods of height measurement options included a portable stadiometer, length board, arm span, crown to heel, and knee height. In the studies of individuals with SB, a stadiometer and linear or segmental measurements were noted in Shurtleff, Walker, Duguay, Peterson, and Cardenas ([37]) and there was no discussion of how height was obtained in the other study (Littlewood et al., [25]). The SCI studies used a wall mounted stadiometer, arm span, supine stadiometer, and supine measurement from head to heel or segmental joint measurements depending on the child's ability to stand (McDonald et al., [27]; Mojtahedi et al., [31]).

Cerebral Palsy

Study Designs and Sample

The five studies primarily used a cross-sectional study design with sample sizes that ranged from 8 to 71 participants. Ages ranged from 2–19 years of age with a mix of male and female participants. Studies included mild to severe function severity based on the gross motor function classification system.

Criterion or Comparison Measures

Some studies included more than a single measurement test. When BIA was used, two studies specified isotope dilution (Bell et al., [4]; Rieken et al., [34]) and one used the DXA scan (Liu et al., [26]). In the four studies using SF, three tested the results against the DXA scan (Gurka et al., [15]; Kuperminc et al., [22]; Liu et al., [26]) and one against isotope dilution (Rieken et al., [34]). A calculated BMI was tested against the DXA scan. An additional test was arm circumference testing versus the DXA scan (Kuperminc et al., [22]).

Findings

Findings for the BIA studies were mixed. The first found the BIA acceptable when using the Fjeld equation at estimating total body water (Bell et al., [4]). Total body water provided percentage BF estimates at the

population level, however, results varied by up to 18% in preschool aged children (Bell et al., [4]). Liu et al. ([26]) found that BIA was moderately correlated to DXA for fat mass and percentage BF highly correlated for fat free mass in a small sample of 8 children. Rieken et al., ([34]) had inaccurate findings for BC when current equations were used with BIA, but had improvement with the development of a new equation that used tibia length versus standing height. In addition, the BIA performed better than SF for BC measurement (Rieken et al., [34]). Two studies created new equations that improved the ability to estimate body fat percentage (Bell et al., [4]; Rieken et al., [34]).

Skinfold measurements had decreased validity and when used with the Slaughter equation underestimated percentage BF when compared to the DXA scan (Gurka et al., [15]; Kuperminc et al., [22]) but overestimated percentage BF when compared to isotope dilution and using the Gurka et al. equation (as cited in Rieken et al., [34]). In one study, SF were not feasibly obtained in 12 of the 61 children (Rieken et al., [34]). When the original equation provided inaccurate results, the researchers developed a new equation (Rieken et al., [34]) or used a correction equation (Gurka et al., [15]). While the new equation performed better, authors identified that further validation is needed. In a small sample size of 8 children, SF had best results for determining percentage BF over BIA methods when compared to DXA (Liu et al., [26]).

Body mass index tests and arm circumferences had a poor correlation with a significant underestimation to percentage BF as measured by DXA (Kuperminc et al., [22]). In addition, arm circumference was not supported when compared to DXA. (Kuperminc et al., [22]).

Limitations

A small sample size and/or decreased generalizability, especially in samples that only consisted of specific subgroups of CP (i.e., preschool-aged children or increased severity of CP) were particularly problematic in these studies. There was a lack of consistency in information provided about the testing methodology used. Additional limitations included the lack of consistency of equations used and development of new equations. While the new equations had improved results, each study identified the need for additional validation testing. When positive findings were found in preschool-aged children, it was noted that while accuracy was seen at the population level, it could not be stated at the individual level.

Spina Bifida

Study designs and sample

One of the two studies reviewed included a pilot study that incorporated a cross-sectional study design with a sample size of 14 children with a mean age of 8.49 ± 3.85 years (Littlewood et al., [25]). The second study performed a retrospective chart review of 709 patients and included 4,968 patient visits from 1965 through 2008 (Shurtleff et al., [37]).

Criterion or Comparison Measure

The first of the two studies compared BIA, with two prediction equations (Cornish et al. and Davies et al. equations), to isotope dilution (Littlewood et al., [25]). The second study did not include a criterion measure, but was included as a seminal study (Shurtleff et al., [37]).

Findings

For the Littlewood et al. ([25]) study, unanticipated results were found. The use of BIA with the prediction equation developed by Cornish et al. (as cited in Littlewood et al., 2003) had the most significant correlation to isotope dilution. This was surprising since the Cornish et al. equation (as cited in Littlewood et al., 2003) had been previously used mainly in adults. An alternative equation, Davies et al. which was previously used primarily in children did not have a strong correlation (as cited in Littlewood et al., [25]). The authors hypothesized that the Davies et al. equation (as cited in Littlewood et al., 2003), which was based on typically developing children,

would not apply to children with SN. In addition the children with SB had additional problems such as growth hormone deficiency, diabetes, and obesity. It was also hypothesized that the children with SB in the study had a fluid status that could have mimicked a healthy adult versus a child which would lead to the adult equation performing better (Littlewood et al., [25]).

The study by Shurtleff and colleagues ([37]) found arm span was acceptable for children with lower level lesions. The study noted that when the children had a higher level of lesion the CDC/NCHS BMI graph underrepresented the child's BMI, thus underestimating obesity in these children. For these children, the authors proposed lowering the graph's BMI percentage by 50% to increase the graph's sensitivity and provide a more accurate reflection of obesity (Shurtleff et al., [37]). The proposed changes provide a unique perspective that provided a cost effective method to better describe the child's obesity levels and highlighted the problems associated with BC measurement in this population.

Limitations

The Littlewood et al. ([25]) authors acknowledged having a small sample size in this pilot study and the need for further study of BC in children with SB. No further evidence or testing was found to support the Shurtleff et al. ([37]) recommendations. Additional evaluation of the suggested BMI percentage changes would be needed to establish the validity of their proposal.

Spinal Cord Injury

Study designs and sample

Two studies met the criteria for review and both used a cross sectional study design. The first sample was 16 highly active athletes (based on the Physical Activity Scale for Individuals with Physical Disabilities [PASIPD]) who all used wheelchairs for ambulation (Mojtahedi et al., [31]). The second was 60 individuals with paraplegia/ tetraplegia and 60 gender, age, and BMI matched controls (McDonald et al., [27]). It should be noted that the study with a sample size of 16 included participants that would be considered adolescent/young adult with an age range of 18-31 with a mean age of 22.0 ± 2.7 for women and 21.9 ± 4.2 for men which met the high end of our inclusion criteria (Mojtahedi et al., [31]).

Criterion measure

The DXA scan was used as the criterion measure for both of the studies with one study comparing SF and BIA measurements and the second comparing BMI.

Findings

Both SF and BIA underrepresented the body fat percentage when compared to the DXA scan (Mojtahedi et al., [31]). Of the two measurements, BIA had body fat percentage readings that were closer to the DXA readings, but had increased variability, total error, and poor correlations to the DXA. When examining the SF measurements with different equations, the Evans et al. equation (as cited in Mojtahedi et al., 2009) using three sites provided the best readings for this highly active, non-ambulating population, but was still inaccurate (Mojtahedi et al., [31]). Further studies that generate new prediction equations for both SF and BIA measurements were recommended for SCI athletes.

In the second study, the tetraplegia group had a significantly lower BMI percentage adjusted for age than their control group and both tetraplegia and paraplegia group had less lean tissue mass and more FM than their control group (McDonald et al., [27]). In general, the traditional BMI criteria were found to significantly underestimate obesity in this population and new proposed criteria were provided that suggested decreasing the BMI cut point for obesity in children and adolescents from \geq 30 kg/m² to 19 kg/m² (McDonald et al., [27]).

Limitations

The small sample size and the inability to obtain the participants' American Spinal Injury Association (ASIA) classification scores were limitations in the first study which could have contributed to the high variability in their findings (Mojtahedi et al., [31]). In the second study, all subjects were college-aged which decreases generalizability of the findings (McDonald et al., [27]). While acknowledging that the DXA is an accepted measure of BC in children, McDonald et al., ([27]) raised potential theoretical concerns about its use as the single criterion measure in children with SCI. They recommended future validation studies with larger samples and a four compartment model of BC measurement.

Discussion

When reviewing the literature over a 10 year period (2003–2013), the lack of studies focused on evidence-based measurement of BC in children with SN is evident. Although a comprehensive search was performed, it is possible pertinent studies were missed. Several studies using BC measurements and subsequent prediction equations were identified, but unfortunately the validity of these methods of measurement were not confirmed.

The level of evidence for the studies in this review reflected a lack of any controlled trials. In addition, the quality of the descriptive studies varied. Many of the studies reviewed used small convenience samples which led to the limitation of decreased generalizability of the findings. The abundance of prediction equations available was evident, but most of the equations had not been generated or evaluated within the SN populations. Multiple studies modified existing equations to fit the population studied, but all would need further validation.

This synthesis supported the potential for inaccuracy of the BMI, SF, and BIA in measuring body fat in the CP, SB, and SCI populations. In the general population where a BMI is recommended as a surrogate for body fat, its accuracy is noted to improve with higher body fat percentage (Centers for Disease Control, [<u>8</u>]). While this was not found in this review, two studies acknowledged the inaccuracies found with the BMI by making adjustments to the cut points or graph to improve its accuracy. In the SB study by Shurtleff et al. ([37]), BMI appeared to be adequate with children who had lower level lesions, but modifications of lowering the BMI percentile curves by 50% for children with higher lesions was recommended. McDonald et al. ([27]) recommended changing the criterion value for obesity in SCI to a BMI of 19kg/m². Both of these findings support the use of BMI in some capacity in children with SN which is favorable due to the accessibility, ease of use and cost-effectiveness of the BMI, but are yet to be validated.

Additional concerns included the lack of consistency of providing information regarding the testing environment, measurement methodology and protocol details such as fasting status, fluid status, or procedures taken with height measurements. With the narrow window of error potentially having a large impact on accuracy of BC measurement, specific procedures for each measurement should be detailed. The final concern is the need for training in obtaining these measures. The potential lack of inter-rater reliability and error is inherent in many measurements of BC. This risk can be exacerbated when working with children and children with SN. Training should include techniques that acknowledge the child's mobility challenges or fear associated with measurements. Without appropriate skilled staff addressing these issues the results can be skewed.

While there are some inconsistencies and measurement challenges reported in the review, there is a consensus that these three populations studied are at a higher risk of obesity. This synthesis highlighted the fact that all indirect measures of BC have a potential for error that increases when they are used with a non-typically developing individual. In the ideal situation, a four compartment model which uses multiple criterion measures could lessen the risk for error but this is often not a practical option. Carefully choosing the best available BC measure for the population that is being assessed, using it thoughtfully and adhering to a standardized protocol for each measure is critical. Options such as modification of existing measurements should be considered (i.e.,

modification of BMI). Additional large population studies need to be performed to create population specific cut points.

Conclusion

Children with SN have many complex health issues and the risk for obesity is often overlooked. In order to work with children and their families to either prevent or treat an abnormal weight status, we must first have an accurate and practical method of identifying when a child is considered overweight or obese. While this is a logical and foundational piece of information, we currently lack evidence-based methods to measure BC or diagnose obesity in this vulnerable population. Acknowledging the specificity that needs to be used in BC measurement methodology and the atypical body morphisms and physiology that are associated with children with SN, it is critical that the scientific community prioritize finding cost effective, reliable, valid, and feasible options for this special population. Until this is done, clinicians, individuals, and families will continue to have a significant barrier in preventing, screening and treating obesity in children with special needs.

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Declaration of Interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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