Anthropometric measurement standardization for a multicenter nutrition survey in children with spinal muscular atrophy

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Abstract
Spinal muscular atrophy (SMA) is a neuromuscular disease associated with nutritional status derangement and altered body composition. New drugs are changing the natural history of the disease, so now more than ever is important to focus on the correct assessment of nutritional status in SMA. We implemented a standardization process for the anthropometric measurement as part of our ongoing longitudinal study of growth patterns in SMA patients. It features a procedural manual, included in this communication, observers training and reliability assessment, of which we publish the values obtained in our pilot study. The standardization process was able to produce inter-observer reliability values in agreement with the literature and a procedure manual is now available for multicentre studies of nutritional status and body composition in SMA and possibly other pediatric neuromuscular disorders.

Introduction
Spinal muscular atrophy (SMA) is a rare and severe neuromuscular disease associated with feeding disorders [1], changes in energy metabolism and impairment of nutritional status [2]. The recent advent of new drugs and clinical trials for SMA are changing the natural history of the disease [1], so now more than ever is important to focus on the correct assessment of nutritional status in SMA patients.

Body composition assessment is paramount to the assessment of nutritional status, and, despite the challenges that it poses, anthropometry is still the most widely used method in the clinical setting. Indeed, there is a lack of literature on standardized procedures suitable for neuromuscular or SMA patients and, due to the rarity of the disease, data collection needs to be carried out by multiple centers and observers, introducing sources of error.

To longitudinally evaluate body composition in SMA children, we started a four center study supported by the Telethon Onlus Foundation (GUP15014), for which we implemented a standardization process for the collection of anthropometric measurement. This communication describes the standardization process and the reliability values obtained.

Materials and methods

Study design
The first step in designing the standardization process was to create a procedural manual for the anthropometric measurements. Based on this manual, a two-day workshop was held to provide a training and standardization setting for the five observers involved in the study. Then, the reliability of the anthropometric measurements was calculated.
The pilot study was approved by the Institution Review Board (Ethic Committee of University of Milan n.7/16) and complies with the Helsinki declaration tenets. Before the study, the parents of the children gave their written informed consent.

Anthropometric manual and anthropometric procedures

The procedures to be included in the manual (Annex A) had to be relevant to the description of body composition and growth of the pediatric patient, but also had to be suitable for use in SMA patients, taking into consideration their reduced muscle mass and tone, muscle contractions, inability to stand, scoliosis, other bone deformities. These procedures are based on those found in other established anthropometric manuals [3–6] and also in approaches previously used in studies of SMA patients [7] or other neuromuscular diseases [8]. Included in the manual are the technical specifications for the instruments needed and the technical procedures to be used for each anthropometric measurement.

Training of anthropometry personnel and error assessment

The first day of the training workshop consisted of a theoretical and practical session in which the techniques included in the manual were presented by the lead anthropometrist and tested by all the observers. The second day was used to assess inter-observer reliability, through the calculation of a complete set of measurements on children affected by SMA, measured by each observer once.

Accuracy was assessed computing coefficient of reliability (R) between each observer and the lead anthropometrist. Observers with a mean R below 0.9 were scheduled for retraining. Inter-observer reliability was assessed computing technical error of measurement (TEM), relative TEM (%TEM), and R values. [9]

Results

Six children (2 boys and 4 girls, 3 healthy and 3 SMA type 2, aged 2–7 years) were measured. After training workshop, 2 of the observers tested displayed acceptable accuracy, whereas 2 required retraining (mean R values for the 4 observers: 0.932, 0.837, 0.915, 0.789). Table 1 shows the inter-observer TEM, %TEM and R for recumbent length, segmental lengths, circumferences and skinfold thickness. Concerning lengths, TEM but not %TEM for recumbent length was higher than TEM for segmental lengths. Arm length had a low R among all measurements. For circumferences, TEMs were lower than 1 cm and R above ~0.9, with arm circumference recording relatively lower values. For skinfold thickness, %TEM was higher compared with other measurements, but R was above ~0.9 except for biceps skinfold which was the worst performer of all measurements.

Discussion

Due to severity and rarity of the disease, a mixed sample of healthy and SMA children was enrolled. In our sample, there were no significant differences in %TEM of the measurement performed (mean for healthy and SMA 9.1% and 7.4%, p-value 0.605), indicating that there was no difference in measurement reliability between healthy and SMA children.

Recumbent length displayed higher TEM than other length measurements but relating measurement error to measurement size through %TEM, recumbent length displayed the lowest value. SMA complicating features prevent the use of a length board to measure recumbent length, thus a composite measure was used, with possible cumulative errors for each segment.

A relative low R value was obtained for arm length, which midpoint is used as a reference point for arm circumference and biceps skinfold. As there were no differences in %TEM values between healthy and SMA children, low R values in these measurements is possibly due to difficulties in the procedure when applied on a supine patient (arm length is measured on the posterior surface of the arm).
We recorded relatively high %TEMs measuring skinfold thickness, but the absolute values of TEMs were still small relative to instrument accuracy (max TEM was 2.6 mm).

Aside from the above reported difficulties, this study shows that high R values can be sought in SMA, but a two-day workshop may be sufficient only in some observers. Concerning the limitations of this study, differences in reliability between healthy and SMA children should be assessed on a larger sample and intra-observer reliability should be assessed in addition to inter-observer reliability. The assessment of nutritional status in SMA patients is currently a topic of interest to evaluate the effects of new therapies on body composition and its role in improving motor function. Anthropometry can be a useful tool for large multicenter studies, but gold standard methods (the 4-compartment model and whole-body magnetic resonance imaging) will also be needed to validate reference and field methods in SMA. Careful consideration of every aspect of the data-collection process should lead the operator to avoid common sources of measurement error and this standardization process could be a useful tool for multicenter studies aiming to fill the gaps in the knowledge of nutritional status of SMA.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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