original article

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Thoracic cirtometry in children with Duchenne muscular dystrophy - expansion of the method

Agenor Garcia Júnior[1], Fátima A. Caromano[1], Adriana M. Contesini[1], Renata Escorcio[1], Lilian A. Y. Fernandes[1], Sílvia M. A. João[2]

ABSTRACT | **Background**: Thoracic cirtometry is a simple and accessible technique to evaluate chest mobility during forced breathing. However, it does not allow for the assessment of compensatory movements commonly used by people with chronic diseases, such Duchenne muscular dystrophy (DMD). DMD is a condition characterized by progressive and irreversible degeneration of the musculoskeletal system. **Objectives**: To expand the method of thoracic cirtometry to allow for the assessment of compensatory movements; to analyze the reliability of the tool; and to describe thoracic mobility of children with DMD during deep breathing. **Method**: Sixty boys, 30 with DMD (10.1±0.5 years) and 30 healthy controls (9.5±0.6 years) participated in the study. The expanded thoracic cirtometry was organized in two phases: 1. the body could move freely, allowing the assessment of compensatory movements (free thoracic cirtometry) and 2. the body without compensatory movements, allowing for the direct study of the movements of the chest (guided thoracic cirtometry). This method includes videotaping and systematic observation of body movements using descriptive and numeric data. We investigated reliability of these measures in both groups. **Results**: Measures of axial and the xiphoid thoracic cirtometry (both free and guided) showed excellent reliability. All measures were significantly different between groups. In DMD boys, free thoracic cirtometry presented a greater value of chest expansion when compared with the guided measures, which probably occurred due to compensatory movements. The most commons were movements of the head, shoulder and torso. **Conclusions**: The expanded thoracic cirtometry method showed excellent reliability and achieved the objectives of determining measures of chest mobility and compensatory movements during deep breath. We suggested its use in the respiratory evaluation of children with DMD.

Keywords: physical therapy; reliability; respiratory assessment; Duchenne muscular dystrophy.

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Introduction

Duchenne Muscular Distrophy (DMD) is the second most common genetic disorder in humans. It is a genetic impairment of recessive nature, with high mutation rates, placed in the Xp21 gene. This mutation causes a flaw in the structure of the muscle membrane, that consequently stops or decrease the production of the protein dystrophin, responsible, among other functions, for the integrity of the basal membrane of the muscle fiber. The clinical presentation of DMD is characterized, mostly, by a progressive and irreversible degeneration of the skeletal musculature, causing incapacity to walk and respiratory insufficiency due to muscle weakness[1].

As a consequence of the muscle weakness of the postural and respiratory musculature, the majority of the patients with DMD develops scoliosis and/or deformities in the thoracic cage, with impairment of the respiratory biomechanics[2,3], reducing the pulmonary volumes and incapacitating thoracic expansion to its maximal capacity and the return to its residual volume[4,5]. As consequence, inappropriate ventilation can be observed with an increased demand from mechanical loads and elevated necessities of ventilation[6].

Several therapeutic approaches, with the purpose to correct the genetic modification are currently under study, but there isn't a cure. Meanwhile, comprehensive, organized and interdisciplinary actions to maintain patients in their best possible physical condition is the best option[1], since it has a preventive function and acts in the primary and secondary aspects of the disease, possibly fostering the natural history and quality of life[7]. The worry with the respiratory function is justified since this

[1]Laboratory of Physical Therapy and Behavior, Department of Physical Therapy, Faculdade de Medicina (FM), Universidade de São Paulo (USP), São Paulo, SP, Brazil
[2]Laboratory of Muscle Skeletal Evaluation, Department of Physical Therapy, FM, USP, São Paulo, SP, Brazil
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system failure is the major cause of death among this population.

In routine assessments and during physical therapy interventions in different respiratory pathologies, the measurement of the amplitude of thoracic movement, through thoracic cirtometry and perimetry, is used because it is practical, fast to administer and have low cost[8]. In situations of forced inspiration, in the presence of muscle weakness, postural muscles can be activated, generating biomechanical compensations[3]. These compensatory movements, in spite of being positive in patients with progressive degenerative diseases, as they provide a strategy to obtain the desired action, are scarcely studied and disregarded during the assessment of the thoracic mobility. Understanding rib cage movements provided by the use of the respiratory muscles and compensatory movements that arise from the use of accessory and postural musculatures, are complementary pieces of information that may help in the understanding of thoracic mobility associated to deep breathing.

Knowing the possible compensatory movements that can support breathing in patients with DMD, in situations where forced breaths are required, can help physical therapists establish strategies with adequately trained motor behaviors.

Thus, the purposes of this study were to broaden the method of thoracic cirtometry to include the assessment of compensatory movements, to analyze the reliability of the tool when administered in healthy children and in those with DMD, and to characterize participant's thoracic movements during deep breath. To do so, the method of expanded thoracic cirtometry was organized in two phases: free body movement during deep breathing, allowing for the assessment of compensatory movements (free thoracic cirtometry) and deep breathing with no compensatory movements, allowing the study of the movements of the rib cage (guided thoracic cirtometry).

Method

This study was approved by the Committee of Ethics and Research of the Medical School of the Universidade de São Paulo (FMUSP), São Paulo, SP, Brazil, under the process number 0685/09.

Subjects

Sixty male children, 30 with DMD (10,1±0,5 years-old) – DMD group – and 30 healthy controls (9,5±0,6 years-old) – H group, participated in this study. The legal guardian of each child signed the free informed consent and children stated that they agreed to participate after demonstration of the procedures, as proposed and approved by the Commission of Ethics and Research of the Institution.

Children were classified as being healthy if they had no history or previous diagnosis of diseases, except communal diseases (such as, otitis, pharingitys or flu), as reported on the interviews performed with the children's guardians. They also had to match the requirements of not having abnormalities in a physical therapeutic postural assessment, administered by a blinded experienced physical therapist.

Children with DMD were assessed by the same evaluator that created a written report on the trunk posture in sitting and on the history of diseases.

The inclusion criteria were the absence of chronic respiratory disease and the capacity to understand the tests proposed. The exclusion criteria were, for both groups, the presence of any acute illness, such as flu and colds or acute muscle skeletal diseases, such as fractures or torsion during the data collection period. This information was collected by the researcher immediately before to data collection.

The present study took place in the Laboratory of physical Therapy and Behavior of the Physical Therapy course of FMUSP, at the Brazilian Association of Muscle Distrophy (ABDIM) and at a State Elementary School of São Paulo City.

Procedures

Cirtometry or thoracic-abdominal perimetry is defined as a set of measurements of thoracic and abdominal circumferences, collected during the performance of respiratory movements[9]. The expression thoracic cirtometry is used when the focus is solely on the thorax movements. This option exists and is commonly performed with people with DMD because these individuals become restricted to the wheelchair during adolescence and the evaluation of abdominal mobility in sitting is questionable since the abdominal musculature is relaxed, facilitating visceral displacement, and the posterior trunk musculature is straightened, limiting the movements in that region. As data collection in the present study was done with participants in a sitting position, thoracic cirtometry was collected.

Thoracic amplitude as measured through cirtometry, is performed with the use of a tape measure, with increments in centimeters, and collected in two regions: on the axilla level and on the level of the xiphoid process. The references used for collection of the measurements in the axilla level were the third intercostal space in the frontal place and the

spinous process of the fifth thoracic vertebrae in the dorsal plane. The references used for collection of the measurement in the height of the xiphoid process were the tip of the xiphoid appendix in the frontal plane and the spinous process of the tenth thoracic vertebrae in the dorsal plane[10].

In recent studies with male young adult volunteers, measurements of thoracic-abdominal cirtometry were shown to be a reliable and useful technique in clinical practice[9-11].

It is suggested that these measurements may be complemented by using the same collection methods but allowing patients to perform head, trunk and arms movements that the patient feel necessary in order to perform deep inspiration and expiration, and thus allowing for the visualization of compensatory movements.

It is proposed that data collection with free movements, named free thoracic cirtometry, should be preceded by the collection with compensatory movements limited (traditional), named guided thoracic cirtometry. With the aim to study compensatory movements, it is proposed that thoracic cirtometry data collection is taped and observed systematically, in such a way that each body segment (head, neck, upper limbs and trunk) can be observed as many times as necessary until the evaluator is able to make conclusion regarding the presence of compensatory movements and describe the type of movement performed. Data must be registered for further analyses.

During the technique of expanded thoracic cirtometry, individuals are oriented to perform a maximal inspiration, while the examiner holds the tape measure, placed around the thorax, over the defined anatomic positions and moves the tape following the thorax expansion. This procedure is performed over the upper and lower levels of the thorax. For each variable, three measures are collected during forced inspiration and expiration, and the difference between them is registered. Therefore, measures collected relate to the thoracic circumference at the inspiratory peak minus the measure collected at the expiratory peak. For research purposes, the largest difference obtained is the one considered in the analysis. To avoid tiredness, a minimum rest is allowed between each collection.

Verbal commands given to patients during data collection should be comprehensible and should reinforce the task. Traditionally, patients are asked not to displace his/her trunk or head forward or backward, and to not move their arms. This action, associated to the blockage imposed by the sitting position, prevents compensatory movements, enabling the isolated evaluation of the mobility of the thoracic cage. During the free cirtometry, that precedes the guided cirtometry, no commands related to the posture or body movements are provided.

Children from both groups were assessed sitting on a wooden bench (standard chair with no backrest), feet flat on the ground (when necessary, a wooden support was positioned) and during the guided cirtometry only, hands were positioned over the thighs.

Data collection

An evaluator that received training for eight hours and assessed ten healthy and ten DMD children performed the measurements of thoracic cirtometry of both groups at baseline and one month after the first collection, providing data for the analysis of intra-rater reliability. Two days after the collection of baseline, another examiner, also previously trained, collected the same measures, generating data for the analysis of inter-rater reliability.

Tests were recorded with a camera placed at an angle of 45° to the subject, at a distance of 2 m from the subject, on a 1m height tripod. The examiner was positioned opposite to the camera to allow observation of the child under assessment.

The recordings were systematic observed and compensatory movements performed during forced expiration and inspiration wre registered.

Data collection and observation of the films were performed by evaluators who had undergone training. Movements of the trunk, shoulder girdle, upper limbs and head were registered and later organized by categories. Thus, each child could present with more than one compensatory movement.

Data analysis

Data normality of the quantitative variables that were collected using a ratio scale were tested using the Kolmogorov-Smirnov test. After verifying normality, the Barlett and Levene tests were performed to check for homogeneity[11]. After the assumptions of normality and homogeneity were confirmed, Pearson correlation tests were used to evaluate the association between the free and the guided thoracic cirtometries at the the axial and the xiphoid levels.

A two-way analysis of variance (ANOVA) was used to compare the free and the guided thoracic cirtometries between groups (DMD and H), with the use of groups and cirtometries measures as factors[12].

Intraclass Correlation Coefficients (ICC) were calculated using the two-way ANOVA results for

reproducibility of data and the one-way ANOVA with repeated measures for calculation of repeatability of the data evaluated. Thus, both between and within group reliability could be calculated[13,14].

To assess the level of reliability between different examiners (inter-rater), the sample was evaluated by two trained examiners, who did not know the results obtained by the other. To analyze the level of reliability of a single examiner (intra-rater), the same subjects were examined in two different occasions, with an interval of one month, so that the examiner did not remember the results[15,16].

For characterization of the subjects and the study of compensations, descriptive analyses were performed.

Results

Characterization of the participants

Demographic characteristics of weight, height and age are listed in Table 1.

Two groups participatns (DMD and healthy controls) of similar age were selected and found to have significantly different weight and height. These results were expected, as DMD affects weight gains and the development of these children. Children with DMD were shorter and heavier. Weight gain in this population may be associated with inactivity[7].

All children with DMD were taking corticosteroid, and none were using antidepressant or analgesic medication. None of the healthy children were taking medications. The use of corticosteroid does not affect the biomechanics and the physiology of respiration.

During the physical assessment of individuals, each dysfunction was categorized in isolation, thus, a child could fit into more than one category of trunk postural dysfunction. It was detected that in the DMD group, 97% of children presented some sort of deformity in the chest, the most obvious being the right bulging (46%) and the barrel chest (23%). In this group, 57% of children had anatomical deformities of the thorax, that were related to the spine, and, of these, 46% had scoliosis on the right, 10% on the left and 1% (only one kid) with scoliosis in "S". These changes made data collection difficult, although it did not prevent it. No chest deformities were found on the healthy group.

Analysis of reliability

Inter-rater and intra-rater reliability of the thoracic cirtometry (axial and xiphoid) were investigated. The reliability in assessing the presence of compensations during forced respiratory movements of children with DMD and healthy controls were also evaluated. As shown in Table 2, all measures studied, for both groups, had excellent reliability demonstrating that they were adequate technical in evaluating thoracic expansibility under the proposed conditions.

Measures of free and guided thoracic cirtometry were performed among children with DMD and healthy controls. All measures of cirtometry were significantly different between the groups, as demonstrated by the p values on Table 3. The same was true for the number of postural compensations, considering each body segment alone.

Thoracic cirtometry of children with DMD were statistically significant different between the free and the guided axial measures, with the free measures being greater than the thoracic expansibility values. As free axial thoracic cirtometry allowed children to perform the compensation as they wished, compensations that emerged to facilitate thoracic expansibility were observed and recorded.

Compensatory movements

The comparison between the number of compensatory movements presented by children was significantly higher in the DMD group with $p<0.001$.

During free cirtometry, it was possible to observe that, in the Group DMD, 13 participants performed shoulder compensations with movements of lifting and external rotation followed by depression and internal rotation; extension followed by head flexion; forward projection with trunk extension followed by return to the initial position; five participants even used a slight hand support as lever for trunk displacement. Other 17 participants used less

Table 1. Characteristics of the subjects according to age, weight and height, and t-test comparison between groups (DMD and H).

	DMD group (n=30)	Group H (n=30)	p value
Age (years)	10.1±0.5	9.5±0.6	0.332
Weight (kg)	41.70±1.20	37.40±1.34	0.009*
Height (cm)	131±2.3	139±3.6	0.003*

Values expressed as mean±SD. *Significant difference ($p<0.05$). DMD: Duchenne muscular dystrophy; H: Healthy.

Table 2. Analysis of the intra-rater and inter-rater reliability of the thoracic cirtometry measures and the compensations of groups DMD and H.

Intra-rater reliability – Group DMD						
Thoracic cirtometry (cm)	**Type**	**First evaluation**	**Second Evaluation**	**ICC**	**Classification**	**P value**
Axial	Free	2.6±1.0	2.5±0.9	0.90	Excellent	0.674
Axial	Guided	1.8±0.8	1.8±0.9	0.96	Excellent	0.599
Xiphoid	Free	2.1±1.1	2.2±0.9	0.95	Excellent	0.632
Xiphoid	Guided	1.7±0.9	1.6±0.8	0.90	Excellent	0.828
Compensation by segment		3.8±0.5	3.8±0.4	0.93	Excellent	0.893
Intra-rater reliability – Group H						
Thoracic cirtometry (cm)	**Type**	**First evaluation**	**Second Evaluation**	**ICC**	**Classification**	**P value**
Axial	Free	5.1±0.9	5.1±0.8	0.99	Excellent	0.948
Axial	Guided	3.8±0.9	3.8±0.9	1.00	Excellent	0.975
Xiphoid	Free	4.7±0.9	4.6±0.8	0.98	Excellent	0.580
Xiphoid	Guided	3.6±0.8	3.6±0.8	1.00	Excellent	0.998
Compensation by segment		1.2±0.5	1.3±0.2	0.96	Excellent	0.893
Inter-rater reliability – Group DMD						
Thoracic cirtometry (cm)		**Examiner 1**	**Examiner 2**	**ICC**	**Classification**	**P value**
Axial free thoracic cirtometry		2.6±1.0	2.4±0.9	0.90	Excellent	0.757
Axial guided thoracic cirtometry		1.8±0.8	2.0±0.4	0.91	Excellent	0.566
Xiphoid free thoracic cirtometry		2.1±1.1	2.2±0.8	0.90	Excellent	0.659
Xifóide guided thoracic cirtometry		1.7±0.9	1.9±0.8	0.90	Excellent	0.622
Compensation by segment		3.8±0.5	3.7±0.8	0.90	Excellent	0.793
Inter-rater reliability – Group H						
Thoracic cirtometry (cm)		**Examiner 1**	**Examiner 2**	**ICC**	**Classification**	**P value**
Axial free thoracic cirtometry		5.2±0.8	5.2±0.8	1.00	Excellent	0.841
Axial guided thoracic cirtometry		4.5±0.7	4.6±0.8	0.98	Excellent	0.568
Xiphoide free thoracic cirtometry		4.6±0.8	4.6±0.8	1.00	Excellent	0.777
Xifóide guided thoracic cirtometry		4.3±0.8	4.5±0.8	0.97	Excellent	0.849
Compensation by segment		1.2±0.5	1.2±0.9	0.94	Excellent	0.901

P<0.05 indicate statistical significant differences between the variables.

Table 3. Comparison of thoracic cirtometry between Duchenne muscular dystrophy (DMD) and Healthy children (H).

Thoracic cirtometry	**DMD**	**Healthy**	*F*	**P value**
Axial free thoracic cirtometry (cm)	2.6±1.0	5.7±1.6	1.4	<0.001
Axial guided thoracic cirtometry (cm)	2.1±1.1	5.4±1.6		<0.001
Xiphoide free thoracic cirtometry (cm)	1.8±0.8	4.5±1.1	0.04	<0.001
Xifóide guided thoracic cirtometry (cm)	1.7±0.9	4.4±1.5		<0.001

P<0.05 indicate statistical significant differences between the variables.

important compensatory movements, associating movements of forward projection with extension followed by return to the initial trunk position associated with head flexion and extension.

In the H Group, 12 participants compensated with flexion and extension movements of the head, four children performed forward projection and return to the trunk starting position, and eight boys performed

compensations with elevation and depression of the shoulders; six participants did associated movements of elevation and depression together with shoulder flexion and extension. All children from group H performed compensations, but of lower impact than those performed by the DMD children.

Discussion

In the evaluation and during physical therapeutic interventions in distinct respiratory pathologies, the measurement of the thoracic range of motion has been used because of its low cost, easy comprehension and administration and appropriateness to the clinical environment. Even when the lungs are not directly involved, as in surgeries of laparotomy, biomechanical changes may occur, and this measurement technique may be used[17-19]. Thoracic cirtometry, in spite of its limitations as a technique, when used with a standardized criteria, can provide the information about the rib cage functional status[9,19,20].

Respiratory assessments of children with DMD provide fundamental data for clinical decision-making, especially in the clinical environment, where physical therapist can take advantage of a thoracic cirtometry examination.

A study with asthmatic patients undergoing osteopathic manipulation demonstrated the effectiveness of the treatment, while using thoracic cirtometry as an outcome measure[20,21]. In children with cystic fibrosis, undergoing physical therapy intervention[22], pulmonary functional status was found to be indirectly reflected in chest expansibility that was obtained by use of thoracic cirtometry. In healthy adults, chest expansibility evaluations through thoracic cirtometry, has been found to be highly reliable and effetive[10]. In this study, all measures of cirtometry in both intra- and inter-rater analyses showed excellent reliability, demonstrating an adequate technical of this measure for the evaluation of thoracic expansibility in healthy and DMD children.

Along with the simultaneous progressive weakness of respiratory muscles, restrictive pulmonary insufficiency, changing the lung expansibility due to postural change also occurs. Most patients with DMD develop a progressive scoliosis when they cease to walk and start to make use of the wheelchair. In this phase, they also develop or accentuate thoracic and pelvic deformities[23,24]. In the characterization and postural assessment of our participants, postural changes were found, with 46% of them showing scoliosis with concavity to the right, 10% to the left and 1% in "S".

In a longitudinal study that evaluated the correlation between thoracic deformities and lung changes, a decreased respiratory vital capacity was found to be associated with the worsening of the thoracic deformities. Deformities and their association with muscle weakness in DMD should be evaluated and monitored during the development of the disease[25].

These progressive postural and motor changes can lead children to build adaptive strategies to achieve the desired movement. In a study to evaluate two-hand function in children with DMD[26], the authors found that as a result of the muscle weakness that is characteristic to the condition, children performed anticipatory postural adjustments to complete their activities.

Respiratory muscle weakness alters lung volumes, impairing lung expansion to its total capacity[25]. Aside from the difficulty to generate lung volume, this loss, associated with muscle weakness, also leads to difficulty in generating protective mechanisms, such cough[27,28]. Progressive respiratory changes with the worsening of the condition, can also lead to adjustments in the ventilatory and kinematics of the respiration, such as the adoption of a predominantly abdominal breathing pattern[29,30].

The present study proposes the expansion of the thoracic cirtometry technique from a guided to a free methods, which could provide information on postural compensations related to the facilitation of breathing in children with DMD. Significant differences were found in all measures of thoracic cirtometry between groups of healthy and DMD children, being the last group characterized by smaller measures. When the values of chest expansibility were analyzed, greater values of free thoracic cirtometry where found when compared to the guided evaluation, only in the DMD group.

To achieve greater thoracic expansibility, participants performed compensations mainly of the head associated with the shoulder and of the thoracic and lumbar spines. Therefore, these children created compensatory strategies, on the same manner that they compensate for other motor functions, with the aim to improve respiratory function. These compensations seem to be beneficial as they allow for an increase in thoracic expansibility.

The limitations of this work are mainly centered at the impossibility to grade compensations performed during the free cirtometry and the absence of an evaluation of abdominal mobility during deep

breathing. Both themes might be objects of future studies from our research group.

Conclusions

The method of assessment tested – expanded thoracic cirtometry – free and guided, with observation of compensatory movements, presented excellent intra- and inter-rater reliability.

Significant differences in the measurements obtained by using the free or guided assessments were found, with the first generating complementary information in the respiratory evaluation of these patients. It was possible to observe that the use of compensatory movements is common and normal during inspiration followed by forced expiration, and these compensations involve a greater number of segments in the DMD group.

The use of the methods of free and guided thoracic cirtometry in routine pulmonary evaluation of children with DMD or similar diseases, such as Becker's Muscular Dystrophy is recommended.

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Correspondence

Fátima Caromano
Universidade de São Paulo
Curso de Fisioterapia da Faculdade de Medicina
Laboratório de Fisioterapia e Comportamento
Rua Cipotânea, 51, Cidade Universitária
CEP 05360-000, São Paulo, SP, Brasil
e-mail: caromano@usp.br