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Reproducibility of the Kids Balance Evaluation Systems Test (Kids-BESTest) and Mini Kids-BESTest for children with Down syndrome

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Abstract:

People with Down syndrome are characterized by change postural control, which makes their balance disturbed. Many therapies focus on the functional improvement of many parameters. In order to properly verify the effects of the methods proposed by therapists, the research tool has to be reliable. This aim of the study was to validate the BESTest and Mini BESTest for people with Down syndrome. The study examined five school-age children. The assessment was performed by the therapist twice. Furthermore, the video from the first test was assessed, and then the second independent expert also assessed the video recording. The results were used for statistical analysis. Excellent reliability was observed for both the Kids-BESTest (ICC 0.95 to 0.97) and Kids-Mini-BESTest (ICC 0.87 to 0.99). The limits of agreement (LoA) in the full form of the test ranged from 0.6 to 2.8; in Kids-Mini-BESTest, it ranged from 0.2 to 0.4. Both Kids-BESTest and the Kids-Mini-BESTest can be successfully used in older children with Down syndrome to evaluate postural control disorders.

Keywords:

postural balance, reactive postural response, genetic disease, balance

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Introduction

Down syndrome (DS) is one of the most common genetic disorders [1], which affects 1 in 605 live births in Poland [2,3]. People with Down syndrome struggle with different consequences of trisomy 21. The authors point to a significant problem with deficits in the postural control system that may provide a partial explanation for functional balance problems [4]. In children with DS, postural responses are slowed down [5], which results in an inadequate response of the central nervous system in postural control during movement to a highly differentiated extent. Furthermore, children with DS have a longer reaction time and deficits of co-contraction of agonist and antagonist muscles [6,7]. Due to the disturbances of the central nervous system occurring in this genetic disorder, the most of people with DS are also characterized by inadequate postural alignment [8], manifesting itself in a faulty posture in these people (knee valgus, pelvic anterior tilt) [9, 10].

Although the majority of children with DS are able to walk, the analysis of various motor patterns reveals inadequate postural control, anticipatory mechanisms associated with the presence of multiple compensations in the peripheral motor system [6,8,9]. Dysfunctions in postural control are often described in DS children and are associated with motor coordination difficulties, problems with sensory-motor integration or simply with clumsy movements [6]. The most likely causes of this phenomenon are exacerbated weakness in the joints, muscle weakness, sensory-motor abilities, cerebellar hypoplasia, and hypertonia [11]. Due to the complexity of the problem of postural control, many aspects forming the concept of stability and balance and the individual differences in the case of people with DS, it seems that the BESTest or mini BESTest are an adequate research tool in the assessment of balance or progress of therapy, as they contain many components that enable the verification of several complex balance processes and a more accurate assessment of the effect of therapy on maintaining balance in specific functional activities such as walking and reaching.

Postural control, viewed as the way the central nervous system regulates and processes sensory information to achieve the adequate postural tension distribution in order to maintain a balanced static and dynamic position [12], represents a system of many complex aspects of human body's control. Therefore, in order to assess the state of postural control, it is necessary to focus on its multiple aspects. Kids-BESTest is used to evaluate *Musculoskeletal components* (muscle strength and joint range of motion) [13], *Sensory systems and Sensory strategies* (vestibular, visual and proprioceptive function and how they are integrated) [14,15], *Anticipatory mechanisms* (dysfunctional feedforward postural adjustments) [16], *Adaptive mechanisms* and *Neuromuscular synergies* (ankle, hip and stepping strategies and feedback postural reactions) [16] and *Internal representations* of stability limitations (reaching in sitting and standing) [17]. Despite its shortened form, the Mini BESTest also contains the most important aspects of postural control: anticipatory, reactive postural control, sensory orientation and analysis of the dynamic gait.

The BESTest and Mini-BESTest are more sensitivity for people with impaired postural body control because they show greater variability in skill levels. Dewar indicated that the proposed tests are better suited for children with reduced postural control, e.g. children with cerebral palsy (CP)[18]. Therefore, the aim of this study is to validate the BESTest and Mini BESTest in another group (which also

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requires control due to the proven inadequate specifity of the neuromuscular control development and associated stability deficits in children with DS).

Material and Methods

Study Design and Participants

Intra-rater, inter-rater and test-retest reproducibility of the Kids-BESTest was examined for 5 school-age children with Down syndrome. Participants were recruited from a local school. The study was conducted in accordance with the guidelines of the Declaration of Helsinki (World Medical Association, 2008).

Children were eligible for inclusion if they (1) had Down syndrome, (2) were aged between 8 and 18 years, and (3) were able to follow child-friendly test instructions. Children were excluded if they had a history of: (1) orthopaedic or neurological surgery within 12 months, (2) uncontrolled seizures or (3) comorbidities interfering in physical functioning e.g. autism, (4) associated cardiovascular condition and loss of functional vision and hearing. Prior to the experiment, children and guardians were provided with written and verbal study information. All guardians signed consent forms and all children signed assent forms.

Outcome measures

Postural control of children with Down syndrome was assessed using the Kids-BESTest according to the protocol published by Dewar et al. [19]. Each of the 36 tasks in the Kids-BESTest was scored from 3 (best performance) to 0 (worst performance) to generate six *Domain scores*, and a *Total Score* ranging from 0 to 108. The tool takes approximately 30 minutes to administer. The subset is designed to quickly identify individuals at risk of falls [20]. It takes 15 minutes to administer and items are scored on a reduced scale from to 2 (best performance) to 0 (worst performance) with a maximum of 28 points. Performance was evaluated by two paediatric physiotherapists (Examiner 1: the first author; Examiner 2: an independent examiner), both having experience in working with children with Down syndrome. To promote consistency, both examiners completed administration and scoring training via the BESTest website and training on the paediatric modifications using the Kids-BESTest protocol.

Procedure

Reproducibility was examined under four conditions: (1) Test-retest real time; (2) Test-retest video; (3) Intra-rater video. To achieve this, children were assessed in real time and all assessments were videoed concurrently using the published Kids-BESTest video recording protocol [18]. Real-time assessments were completed on Day 1 (n=5) and Day 2 (n=5) by Examiner 1.The interval between real-time assessments was 1 to 10 days. Video-based assessments were performed retrospectively after all real-time assessments were completed. Test-retest reproducibility was evaluated from Day 1 and Day 2 performance in real time and via video by Examiner 1. Intra-rater reproducibility was assessed with Day 1 video by Examiner 1. Inter-rater reproducibility was assessed with Day 1 video by Examiner 1 and separately by the independent Examiner 2. In each case, reproducibility was evaluated for the Total Score and all Domains of the Kids-BESTest (6 domains) and the Mini-BESTest (4 domains). In each case, assessments were conducted in an open room space with equipment and floor markings used according to the Kids-BESTest administration and video protocols. [19].

Statistical analysis

Reproducibility is the degree to which repeated measures of the tests

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provide similar results. Agreement assesses how close the results of repeated measures are and the margins that may be used to represent real clinical change, as opposed to random measurement error. Reliability evaluates how well children can be distinguished from one another despite measurement error. Statistical analysis was performed using Statistica 13.1 software package (StatSoft).

Limits of agreement (LoA) were evaluated using the Bland-Altman method (20). Suitable percentage agreement was set *a priori,* consistent with the previous study of typically developing children. For the Kids-BESTest, Total score was defined as: excellent = >90% within 4 points, good = >80% within 4 points, fair = >60% within 4 points and poor = <60% within 4 points. For the Kids-Mini-BESTest, Total score was defined as: excellent = >90% within 2 points, good = >80% within 2 points, fair = >60% within 2 points and poor = <60% within 2 points. For the domains the *a priori* agreement values were set at 2 points for Kids-BESTest domain scores and 1 point for Kids-Mini-BESTest scores. The standard error of measurement (SEM) was calculated to indicate the error of measurement of both tools. The LoA was calculated as the range within which different examiners or the same examiner produced similar scores on separate assessment occasions. Reliability was calculated based on intra-class correlation coefficients (ICC). Consistent with previous work [19, 21], an ICC was defined as excellent = > 0.75; good = 0.74 – 0.60; fair = 0.59 – 0.40; and poor = < 0.4.

Results

The study examined 10 children with Down syndrome. Five people were excluded at the initial stage of research (3 - intellectual disabilities, 1 - colds, 1 - lack of willingness to cooperate). The included participants were between 12 and 17 years old and were able to complete all components of the Kids-BESTest and Kids -Mini-BESTest. Detailed characterization of the group is presented in Tab. 1.

Table 1. Baseline characteristics

Characteristics	Group
Gender	3 (girls) 2 (boys)
Age (years)	14.4
Height (cm)	150
Weight (kg)	54.4
BMI	24.18

Kids-BESTest results

The results of the extended test formula show that the largest intra-rater agreement between one researcher occurs for *Stability limits* (ICC 0.97- excellent, see Tab.2) and the smallest - for *Biomechanical constraints* (ICC 0.53-fair, see Tab. 2). A comparison of the assessment of both researchers was obtained for *Reactive postural response* (ICC 0.98) and *Stability limits* (ICC 0.97, see Tab.2). A poor result (ICC 0.22) was found for *Biomechanical constraints.* Generally, the intra-rater reliability of Kids -BESTest all Domains od was also excellent (ICC 0,95-0,97 –Tab. 2).

However, the video assessment was the least differentiated in individual test components, with the ICC results ranging from 0.78 to 0.89. The highest correlation

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Table 2. Reliability analyses for A. Kids-BESTest and B. Kids-Mini-BESTest

TEST	Dominates	Intra-rater n=5, one assessor		Inter- rater two assessor (n=5)		Test-retest video (one assesor)	
		ICC	SEM	ICC	SEM	ICC	SEM
Kids BES	Biomechanical constraints	0.533	1.14	0.22	0.93	0.8	0.37
	Stability limits	0.97	0.36	0.97	0.42	0.88	0.85
	Transitions	0.83	0.49	0.56	1.36	0.85	0.55
	Reactive postur-al response	0.87	0.87	0.98	0.33	0.87	0.72
	Sensory orienta-tion	0.96	0.35	0.92	0.61	0.78	0.80
	Stability in gait	0.53	1.23	0.91	0.45	0.89	0.54
	Total	0.95	1.46	0.97	1.22	0.97	1.18
Kids Mini-BES	Anticipatory	0.9	0.31	0.7	0.38	0.89	0.31
	Reactive postur-al control	0.85	0.37	0.65	0.56	0.89	0.31
	Sensory orienta-tion	0.4	0.4	0.4	0.4	0.45	0.38
	Dynamic gait	0.81	0.47	0.95	0.3	0.94	0.31
	Total	0.9	0.78	0.87	0.89	0.99	0.27

between the three tests occurred in *Stability limit*, where the ICC value ranged between 0.88 and 0.97.

Kids-Mini-BESTest Results

The results of the shortened formula show that the largest intra-rater agreement between one researcher occurs for *Anticipatory* (ICC 0.90- excellent, see Tab.2), whereas the smallest - for *Sensory orientation* (ICC 0.4-fair, Tab. 2). The comparison of the assessment of both examiners had the highest agreement in the *Dynamic gait* test (ICC 0.95). A fair result (ICC 0.4) was obtained for *Sensory orientation*, where tests of two individuals were analysed. Simiraly to Kids-BESTest, the intra-rater reliability of Kids -Mini-BESTest all Domains was also excellent (ICC 0,87-0,99 – Tab. 2). In this case, the video assessment by the examiners was the parameter with the highest agreement (ICC 0.99). The lowest value in video assessment was found for *Sensory orientation* (ICC 0.45). The highest correlation between the three tests was found in *Dynamic gait*, where the ICC value ranged from 0.81 to 0.95.

Analyzes of LoA for Kids-BESTest demonstrated that the systematic error (bias) hovers from 0,6 to 2,8. In the case of the shortened formula, however, the values were much smaller and ranged from 0.2 to 0.4. Detailed results are presented in Tab.3.

Discussion

Kids-BESTest and Kids-Mini-BESTest cover many aspects of

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Table 3. Summary agreement (LoA) for BESTest and MiniBESTest

	Kids BESTest			Kids Mini- BESTest		
	Lower Agreement Limit	Upper Agreement Limit	MoD (bias)	Lower Agreement Limit	Upper Agreement Limit	MoD (bias)
Intra-rater n=5, one assessor	-5.84	7.04	0.6	-3.02	3.42	0.2
Intra-rater n=5, two assessors	-4.71	10.3	2.8	-2.22	3.03	0.4
Test-retest video (one assesor)	-7.19	9.19	1.0	-0.67	1.07	0.2

Legend: MoD - magnitude of difference

postural control and can therefore be used to evaluate complex motor problem in people with Down syndrome and the effects of the therapy dedicated to changes in motor control. Our results show that the tests can be successfully performed and reproduced in some children with Down syndrome.

Unfortunately, as shown in our study, this is a demanding research group, and in order to fully perform the tests correctly, the participants have to cooperate, understand the instructions and be willing to follow them, which in the case of more severe intellectual disabilities, which are not uncommon in people with Down syndrome, can make the study difficult or impossible. The study revealed that the above-mentioned tests help detect clinical changes (high agreement) and changes in the body's postural control.

The results of studies of children with Down syndrome indicate that the Kids -BESTest is a more sensitive test when assessing motor control disorders compared to the Mini-BESTest. This is probably due to the more detailed nature of the study and a greater number of trials that differentiate postural control problems in children. These results have been documented in studies of children with typical school-age development [19]. A similar relationship has also been observed in patients with neurological dysfunctions in both children (CP) [18] and adults (stroke) [22]. However, despite greater test reliability, the examinations of children with Down syndrome found a rapid decline in the concentration and attention during the tests. Therefore, performing the full version of the test in younger participants may be difficult.

Dewar et al. [18,19] suggest that the Kids-BESTest may be more effective in differentiating postural control in children with CP than in children with normal development. This is because the results for some domains have reached a threshold of children with typical development, which lowered the reliability score for this population [19]. However, none of the children with CP [18] nor most of those with Down syndrome reached the level of any part of the study. The group of children not developing typically in the course of CP and Down

syndrome showed greater variability in skill levels, enabling the test to effectively differentiate children in this group [23].

The differences in examination procedures (two examiners, two examinations performed by one examiner, video recording) allowed to assess how best to conduct the tests. Although the results did not show high differences between the methods, the best result of the tests was obtained in the case of the video assessment of both the short and the full version of the test (ICC 0.97 and 0.99). In the case of one examiner, the results were: ICC 0.96 and 0.9. This difference may occur because some parts of the test require the examiner to respond to minimal changes such as the lateral inclination of the hips, body trunk or even foot movements. Undoubtedly, it is much easier to see these subtle changes during the assessment of video recordings. Therefore, in order to achieve the best results, we recommend combining real-time scoring with a retrospective review of video recordings to confirm scoring in children with Down syndrome. This is consistent with the Kids-BESTest recommendation for typically developing children [18,19].

The lack of literature showing the possibility of the assessment of the complex postural control system in children with Down syndrome without the use of specialized equipment, the lack of attempts to validate Kids BESTest and Mini BESTest in this group of people, and the indications of the clinical usefulness of the tests (e.g. to assess the effects of the therapy) led to the attempt to write this paper. We are aware of many limitations of the study, but it represents a starting point for further research.

The main limitation of the study is small group size. Initially, a group of 10 participants with Down Syndrome was included in the study, but during the research, several children were excluded due to the absence from school and several were unable to complete more than 3-4 attempts with full concentration as their attention was distracted. Furthermore, the children showed a lack of willingness to continue the test although the Kids BESTest test is composed of several components.

In our opinion, performing the above test in children with Down syndrome is difficult but there are no complications. It cannot be applied to the entire study population but only to a group of children who are willing to cooperate. However, the strength of this study is its reliability by critical analyze.

In conclusion, both Kids-BESTest and the Kids-Mini-BESTest can be successfully used in older children with Down syndrome to assess postural control disorders. Both tests can effectively assess balance and its changes in children with DS. Only people with mild intellectual disabilities or normal intellectual development can be included in the tests.

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