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ORIGINAL ARTICLE

Turkish version of Brief Ataxia Rating Scale

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ABSTRACT

Aim: Our aim was to perform the Turkish-language adaptation of a practical ataxia rating scale for children.

Methods: The Brief Ataxia Rating Scale was subjected to cultural adaptation following receipt of the requisite permissions. Thirty-six children aged 4–18 years followed-up with a diagnosis of ataxia were included in the study. Evaluation of each child was recorded on video. The video recordings were scored independently by nine observers (four physiotherapists, one pediatric neurologist, and four pediatricians). Intra-rater reliability was tested by the same video images being scored twice, at 15-day intervals, by a pediatric neurologist. Intraclass correlation coefficients were used for inter-rater and intra-rater reliability. The Scale for the Assessment and Rating of Ataxia was used for concurrent validity.

Results: Good to excellent reliability was determined among the nine observers in terms of total scores with the intraclass correlation coefficient among the nine observers (intraclass correlation coefficient = 0.926; 95% CI: 0.885–0.956). Intra-rater reliability analysis results exhibited strong reliability in terms of scores elicited at two-week intervals (intraclass correlation coefficient = 0.967; 95% CI: 0.890–0.987, $r = 0.97$, $p < 0.001$). At concurrent validity analysis, a strong relation was determined between total Scale of the Assessment and Rating of Ataxia score and total Brief Ataxia Rating Scale score ($r = 0.942$, $p < 0.001$).

Conclusion: The Turkish-language adaptation of the Brief Ataxia Rating Scale is reliable and valid for application in children.

➤ IMPLICATIONS FOR REHABILITATION

- This study shows the reliability and validity of the Turkish language adaptation of brief ataxia rating scale in children.
- The scale being both practical and easily applicable to ataxic children will contribute to broadening its use in the pediatric age group in particular.

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Brief Ataxia Rating Scale; Turkish-language adaptation; children; reliability; validity

Introduction

Ataxia is a disorder of balance and coordination. Conditions accompanying ataxia include coordination weakness, imbalance, dysarthric speech, nystagmus, and swallowing disorder. The reported incidence of acute cerebellar ataxia in children is 1/100 000–1/500 000 [1,2]. Acute ataxia is the most common type, representing 40% of all cases as acute post-infectious cerebellar ataxia. Toxic causes are another very common source of acute childhood ataxias, accounting for approximately a further 30–32% [2–4]. The most common causes of childhood acute ataxia are acute post-infectious cerebellar ataxia, intoxications and acute disseminated encephalomyelitis. Other causes include cerebellar neoplasms, acute hydrocephalus, acute cerebellitis, traumatic or vascular causes, paraneoplastic causes and autoimmune diseases [3]. Congenital abnormalities, degenerative diseases, and hereditary ataxias are involved in the etiology of chronic and progressive ataxia. The estimated prevalence of acquired, hereditary and mixed childhood ataxia is 26/100 000 among children in Europe [5].

The International Cooperative Ataxia Rating Scale (ICARS)' for the evaluation and rating of ataxia was first developed in 1997 [6]. The Scale for the Rating and Assessment of Ataxia (SARA), which assessed various parameters of cerebellar ataxia, was subsequently developed as an alternative to ICARS by Schmitz-Hübsch et al. [7] in 2017 through a multicenter study in Europe, and was shown to be capable of use in children [8]. The Brief Ataxia Rating Scale (BARS) was developed by Schmahmann et al. in 2009 [9] following a more extensive study of a modified form of ICARS. BARS contains five headings and is an effective and practical measurement tool [9].

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Supplemental data for this article can be accessed here.

The Turkish-language reliability and validity of BARS has not previously been investigated in either pediatric or adult cases in Turkey. Children's attention and motivation spans are more limited than those of adults. In particular, physical or partial mental disabilities may be present in pediatric ataxia patients. The use of an effective and also a practical scale is, therefore, an important requirement in terms of meeting these needs. The purpose of this study was to test the validity and reliability of the Turkish-language version of BARS, a practical ataxia rating scale, in children.

Methods

Patients and study design

Approval for this descriptive study was granted by the Karadeniz Technical University Scientific Research Ethical Committee (Number: 86, Date: 16 April 2018).

Thirty-six children aged 4–18 years, presenting to the Karadeniz Technical University Farabi Hospital pediatric neurology clinic between April 2018 and May 2019 and followed-up with a diagnosis of ataxia were included in the study. Patients with severe cognitive disability were excluded. Nine cases from among the 49 presenting to our clinic during the specified period were excluded for not being within the study age group (seven cases under four and two over 18), and four could not be tested due to severe cognitive disability. All the remaining patients agreed to participate. Written and verbal consent was obtained from families, and consent was also obtained from children aged 12 or over. The demographic data of the patients enrolled were recorded. All observers, and the clinicians completing the translation from the original article, received pre-information instruction (by EAA, and AE, [the first and second authors of the present paper]) in the form of a number of sessions regarding the study content, video observation, and the scoring system employed. Video recordings were subsequently taken of all patients for each test item. The video recordings were then scored independently by nine observers, four physiotherapists (with 5–13 years' experience in the field), a pediatric neurologist (with 10 years' experience), and four pediatricians (with 6–10 years' experience), three of whom were working in the field of pediatric neurology. "EAA" also scored the same video images twice, at 15-day intervals.

BARS consists of five clinically important functional domains of ataxia: (1) Gait (0–8 points), (2) Knee-tibia test (right and left, 0–8 points), (3) Finger to nose test (right and left, 0–8 points), (4) Dysarthria (0–4 points). (5) Oculomotor abnormalities (0–2 points). Possible scores range between 0 and 30. Items 2–3 are related to limb kinetic function and are rated bilaterally; the mean scores for both sides are added to the total score [9].

The Scale for the Assessment and Rating of Ataxia (SARA) is a 40-item ataxia evaluation and rating scale consisting of eight main headings. The Turkish version, SARA-T, consists of eight categories examining (1) Gait (0–8), (2) Stance (0–6), (3) Sitting (0–4), (4) Speech disturbance (0–6), (5) Finger chase (0–4), (6) Nose-to-finger test (0–4), (7) Fast alternative hand movement (0–4), and (8) Heel–shin slide (0–4). Items 5–8 are related to limb kinetic function and are rated bilaterally; the mean scores for both sides are added to the total score [7]. Total scores range from 0 to 40.

Translation of BARS into Turkish

Permission was obtained from the corresponding author of the study team responsible for developing the original scale [9]. The standard World Health Organization protocol regarding the adaptation of scales into other languages was applied in this adaptation of BARS into Turkish [10]. The scale was translated independently into Turkish by a native Turkish-speaking pediatric neurologist, a native Turkish-speaking physiotherapist, and a native English-speaking translator (EAA, AE, and NS [a native English-speaker]). The final form of the scale was reached on the basis of consensus at several subsequent meetings. "NS" subsequently translated the scale back into English. The text was then sent to the corresponding author of the team [9] that developed the scale for comments and suggestions. Final approval was received. Following amendment, the scale was translated back into Turkish. The Turkish-language form was finally reproduced for each observer (Supplementary Appendix 1).

Statistical analysis

Data analysis: The data obtained were analyzed on IBM SPSS (Statistical Package for the Social Sciences) 23.0 software (SPSS, Chicago, IL). p values <0.05 were regarded as significant. Categorical variables were expressed as number and percentage, and interval variables as mean ± standard deviation. Reliability was tested using inter-rater and intra-rater reliability analyses. Intraclass correlation coefficient (ICC) values with 95% confidence intervals and "Cronbach's alpha" were used to evaluate the internal consistency of the total BARS. Pearson's correlation coefficient was used for concurrent validity.

Inter-rater reliability analysis: Measurements were recorded using a video system. The video recordings were scored independently by nine different observers. Since the scale is a numerical measurement, we investigated intraclass correlation (ICC) among the nine raters' scores. Inter-rater reliability was tested using the ICC in a two-way random effects model, with absolute agreement, values greater than 0.90 being regarded as excellent reliability, between 0.75 and 0.90 as good reliability, between 0.50 and 0.75 as moderate reliability, and less than 0.50 as poor reliability [11]. Intraclass correlations were also evaluated for subtests (gait, dysarthria, knee-tibia test, finger-to-nose test, and oculomotor abnormalities) among the nine observers.

Intra-rater reliability: A single observer's measurements were assessed at two-week intervals for the same video recording, and the results of both were recorded. Intra-rater reliability was tested using the ICC in a two-way random effects model, with absolute agreement for a single rater. Agreement between ICC and scores was examined. Relations between two analyses were also examined using Pearson correlation analysis.

Validity: SARA was used for concurrent validity. The BARS and SARA scoring tests were compared using Pearson's correlation coefficient. SARA was used to test validity. Since there is no Turkish-language version of SARA in the literature, the requisite permission was obtained from the paper's "corresponding author" [12].

Results

Thirty-six children were included in the study, 14 (38.9%) boys and 22 (61.1%) girls, with a mean age of 10.5 ± 3.85 years. Ataxia telangiectasia was the most common diagnosis ($n = 9$, 25.0%). Demographic data and diagnoses are shown in Table 1. BARS scores for each observer are presented in Table 2, and diagnoses are shown in Supplementary Appendix 1.

Inter-rater reliability analysis: The intraclass correlation coefficient also exhibited good to excellent reliability among the nine observers (ICC = 0.926, 95% CI: 0.885–0.956). Reliability analysis for each category among the observers revealed that ICC values

Table 1. Patient characteristics.

Features	Total ($n = 36$) Mean (SD) (min–max)
Age (years)	10.5 (3.85) (4–18)
Body height (cm)	134.22 (21.05) (90–190)
Z^a score	−0.88 (1.38) [(−3.66) to (+2.75)]
Body weight (kg)	34.91 (14.73) (12–67)
Z^a score	−0.48 (2.13) [(−8.15) to (+2.89)]
BARS (total score)	5.412 (7.22) (1–18)
SARA (total score)	11.69 (8.45) (1–30)

SD: standard deviation; *n*: number of patients.
[a]Calculated according to Centers for Disease Control and Prevention.

Table 2. Observers' descriptive statistics of the brief ataxia rating scale.[a]

Rater	Mean (standard deviation)	Min–max
R1	6.24 (5.68)	0–18
R2	7.50 (5.57)	0–20
R3	7.53 (5.69)	1–20
R4	7.12 (5.42)	0–20
R5	7.86 (5.77)	1–19
R6	7.49 (5.48)	1–19
R7	6.72 (5.06)	0–17
R8	7.49 (5.53)	1–19
R9	7.04 (5.97)	0–18

[a]Intraclass correlation coefficient for inter-rater agreement is 0.926 (95% CI: 0.885–0.956).
Internal consistency of the scale is 0.992 (95% CI: 0.987–0.995).

Table 3. ICC values of interobserver agreement on BARS subdimensions.

Subdimensions of BARS	ICC	95% CI
Gait	0.924	0.885–0.955
Knee-tibia test	0.614	0.491–0.740
Finger to nose test	0.770	0.668–0.857
Dysarthria	0.863	0.798–0.917
Oculomotor abnormalities	0.739	0.634–0.835

ICC: intraclass correlation coefficient; CI: confidence interval; BARS: Brief Ataxia Rating Scale.

exhibited good to excellent reliability (≥0.75) in gait and dysarthria and the finger-to-nose test, and moderate reliability in the knee-tibia test and oculomotor anomalies (0.50–0.75) (Table 3).

Intra-rater reliability analysis: Intra-rater reliability from the same video recordings scored by one observer at two-week intervals also exhibited strong reliability (ICC = 0.967, 95% CI: 0.890–0.987). The relation between the two values also exhibited strong correlation ($r = 0.97$, $p < 0.001$).

Internal consistency: Cronbach's alpha coefficient for the internal consistency of the five-item scale was 0.992 (95% CI: 0.987–0.995). This showed that the Turkish-language adaptation of the scale exhibited a good level of consistency.

Concurrent validity analysis: The correlation between the total SARA test score used for concurrent validity and total BARS score was strong ($r = 0.942$; $p < 0.001$) (Figure 1).

Discussion

Our study represents the first adaptation of BARS into Turkish. Reliability of the BARS test (ICC) was determined among the nine observers (ICC = 0.926, 95% CI: 0.885–0.956). The intra-observer reliability value for the single observer's same video recordings was 0.967 (95% CI: 0.890–0.987). An r value (correlation coefficient) of 0.942 was determined at concurrent validity analysis calculation by comparison with the SARA test ($p < 0.001$). Good to excellent reliability and validity were determined in the children in this study.

There are no ataxia rating scales with proven reliability and validity in Turkey, and none that have been adapted into Turkish for children. BARS permits evaluation of four motor areas – gait, kinetics, speech and oculomotor functions. The first validation of an ataxia rating scale from Turkey was performed in 2017. This examined the effectiveness of ICARS and SARS in adult multiple sclerosis patients. This test was subjected to cultural adaptation and was found to be valid and reliable [12]. The validity and reliability of the more up-to-date BARS have not previously been investigated. From that perspective, our study yielded novel findings.

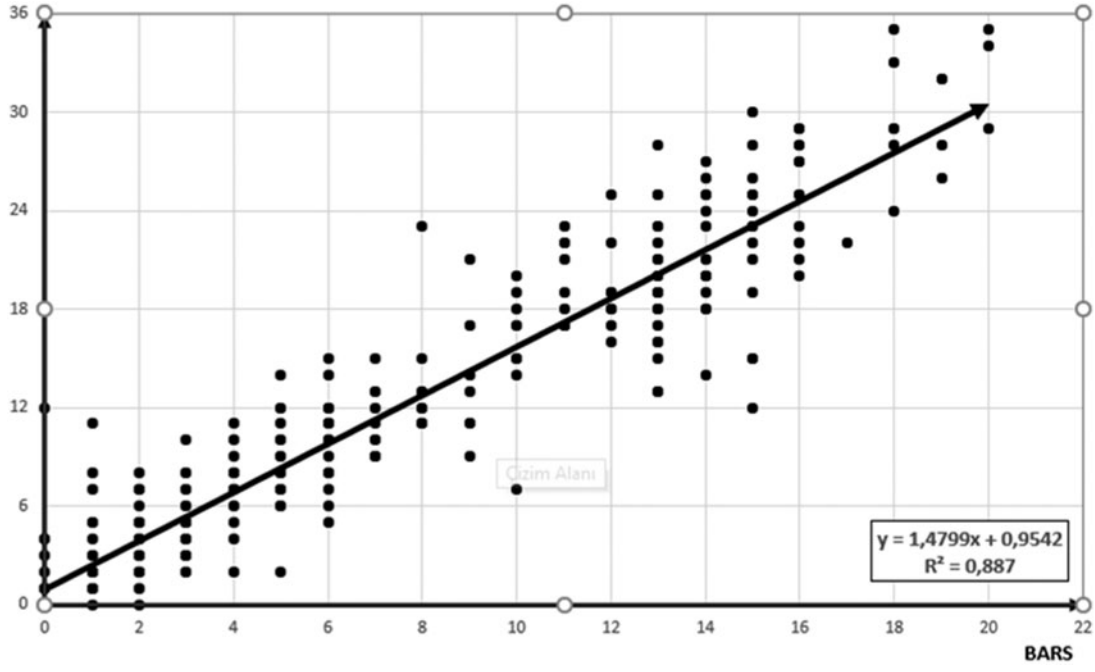

Figure 1. Correlation between total BARS and SARA scores.

Thirty-five cases were evaluated in a previous adaptation of BARS into Brazilian Portuguese. As noted in that study, the ICARS and SARA evaluation process involves longer tests compared to BARS. BARS is easy to learn and effective in the context of cerebellar motor functions [13]. It is also effective in pediatric patients as a practical and reliable test. Similarly, it has also been shown to be reliable in early onset ataxia [14]. One study involving healthy children (4–16 years) showed that BARS is reliable for pediatric patients among the different ataxia rating scales (ICARS, SARA) [15]. BARS is an important rating scale, not only for hereditary ataxias, but also in terms of etiologies of other ataxias, such as autoimmune diseases, vasculopathy, multiple system atrophy, and alcoholic cerebellar degeneration [13,16]. Its efficacy has also been investigated in pediatric patients with brain tumors in recent years, and the test has been found to be reliable and valid [17]. Application of BARS items is also reported to be easy in children with neurodegenerative ataxia and ataxic cerebral palsy [18].

ICC values in ataxia scales in adults are high. However, this information is "incomplete" for children, because coordinated motor movements in children are age-dependent. Physiologically, immature motor characteristics may "overlap" with "ataxic" features. Brandsma et al. [15] reported that children achieved the optimal adult score for BARS at approximately 11 years of age. ICC values were used for inter-rater reliability analysis in the present study. The results of that analysis showed good to excellent reliability among the nine observers' total BARS scores (ICC = 0.926). BARS was previously adapted into Brazilian Portuguese in adult patients, with a BARS ICC of 0.94 being determined [13]. Brandsma et al. [14] reported an ICC value of 0.966 for inter-rater reliability and an ICC value of 0.913 for intra-rater reliability analysis for BARS in early onset ataxias. In our study, ICC values were 0.926 for inter-rater reliability analysis and 0.967 for intra-rater reliability analysis. In terms of sub-tests, the highest ICC value was reported for "gait" (ICC values 0.958 in gait, 0.782 in kinetics, 0.807 in speech, and 0.705 in oculomotor functions) [14]. The highest ICC value in our study was also determined for the "gait" subdimension (ICC = 0.924). Studies involving BARS are limited, particularly in the pediatric age group [14,15,18,19]. We think that more accurate comparison will be possible as more reliability studies are performed in the pediatric age group.

Concurrent validity analysis in our study was performed using the SARA scale, previously the subject of cultural adaptation into Japanese [20], Brazilian Portuguese [21], Chinese [22], and most recently Turkish in 2017 [12]. Lawerman et al. determined an ICC value of 0.63 in total SARA scores in a European cohort aged 4–16 years. That study reported that pediatric scores after the age of 11 were close to adult outcomes [14]. The authors also reported that reliability was more significant after the age of eight. A large proportion of our study group consisted of patients aged eight or more ($n = 30/36$, two children aged four ($n = 2$); three aged five ($n = 3$), and one aged six ($n = 1$)). Good to excellent "interobserver reliability" was determined in our study, and from that perspective our findings are compatible with the previous literature. The five-item BARS can also be administered more quickly than the eight-item SARA test. Depending on the specific status and age of the child, estimated test times are approximately 10 min for BARS, 15-20 min for SARA, and 30 min for ICARS. In the absence of severe test-limiting comorbidity, BARS and SARA are generally applicable in children older than four years of age [23]. We, therefore, employed the SARA scale for comparative purposes at BARS reliability analysis due to its ease of application, particularly in children. We determined high correlation, indicating BARS validity.

Specialists from various branches encountering ataxia patients were included in a multidisciplinary manner in the present study. Response to treatment or progression of disease in children with ataxia can be monitored through ataxia rating, which permits objective evaluation (ICC = 0.967; $r = 0.97$, $p < 0.001$). Such patients may be encountered by pediatricians in the pediatric emergency department, as well as by physiotherapists and pediatric neurologists. Studies concerning the effectiveness of physiotherapy or device-accompanied physiotherapy in adult ataxic cases have become increasingly important in recent years [24–28]. It is important for scoring systems in the pediatric age group to be available, and for them to be disseminated more widely through cultural adaptation. Chronic cases are frequently assessed by pediatric neurologists, while pediatricians in the emergency department also directly encounter acute cases. BARS being both practical and easily applied to children will further encourage the further dissemination of the scale.

Strengths of the present study include a larger number of observers compared to other research in the literature [10,12–15,20], the fact that each case was evaluated independently from video records, and that the study involved specialists from several disciplines, such as physiotherapy, pediatrics, and pediatric neurology. Another important feature is the inclusion of children with ataxia, a rare disease group.

Our study is important in showing the reliability and validity of the Turkish-language adaptation of BARS in children and is the first such research conducted in Turkish and from Turkey. The scale being both practical and easily applicable to ataxic children will contribute to broadening its use and to increasing the number and quality of studies in the pediatric age group in particular.

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Ethical approval

The approval for this study was granted by the Karadeniz Technical University Scientific Research Ethical Committee (No. 2018/86, dated 16 April, 2018).

Disclosure statement

The authors report no conflicts of interest and certify that no funding has been received for this study and/or preparation of this manuscript.

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