CONSTRUCT VALIDITY OF INDONESIAN LANGUAGE VERSION OF CHILDHOOD AUTISM RATING SCALE™ SECOND EDITION

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ABSTRACT

Combination of The Childhood Autism Rating Scale™ Second Edition - Standard Clinical Tool (CARS2-ST) with criteria for autism spectrum disorder (ASD) based on Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5) is thought to improve diagnostic process. To meet diagnostic needs, localized, Indonesian-translated version of CARS2-ST is developed. Therefore, assessment of construct validity of the Indonesian-language translated CARS2-ST must be performed. A cross-sectional study was conducted in Child Development Center in Surabaya, Indonesia from August to December 2019. Diagnosis of ASD then performed by using CARS2-ST and DSM-5 criteria. To assess construct validity, principal components analysis and Kaiser-varimax rotation was performed for CARS2-ST in order to determine factors. DSM-5 criteria for ASD was used to compare scale. There were 201 children aged 2 - 6 years old with reported speech and behavior problems. Sixty-six children were diagnosed for ASD. Factor analysis using Kaiser-varimax rotation indicates a significant two factors: social communication and interaction factor and restrictive stereotyped behaviors and sensory factor sensitivities. The factors satisfyingly reflects the criteria for ASD in DSM-5. Construct validity of Indonesian-translated version of CARS2-ST is confirmed, as shown by its correspondence with DSM-5 criteria for diagnosing. This study supports the continued relevance of the Indonesian CARS2-ST in ASD assessment.

Keywords: ASD assessment, Diagnostic criteria, DSM-5

INTRODUCTION

Autism spectrum disorder (ASD) is a group of neurodevelopmental disorders characterized by a deficit of social communication and interaction and the presence of limited or repetitive behaviors or interests.¹ According Center for Disease Control and Prevention (CDC), the incidence of autism continues to increase over the last few decades. In 2014, it was found that the prevalence rate of ASD had risen to 1 per 59 children.² Diagnosis of ASD in childhood is often challenging due to the lack of gold measurement standard and various aspects that must be assessed.³ Moreover, lack of research on the validation of tools for screening and diagnosis in low- and middle-income countries creates problems for the development of evidence-based diagnostic tools according to local community conditions.⁴ Some other factors that influences this undervalidation are inaccessible health facilities, the inability of diagnosis bt health workers, and inadequate diagnostic instruments.⁵

Childhood Autism Rating Scale (CARS) was developed for the differential diagnosis of autism from other developmental disorders. In 2010, Schopler released the CARS, Second Edition (CARS2), which includes a Standard version as clinical tool for diagnosis (CARS2-ST). CARS2-ST is used for children aged 2-6 years or with an IQ score of less than 80.⁶ CARS2-ST has several advantages over other instruments, including being easy to use, having broad diagnostic criteria; including communication disorders and social interactions, maladaptive behavior, applicable to children aged 2-6 years, uses objective and measurable scores, and is a standard instrument which is accepted globally. These advantages make CARS2-ST has a good psychometric value.⁶⁻⁹ In the other hand, DSM-5 uses criterion for diagnosing mental disorder in childhood based on statistically available evidence. Although it is a common method for clinical diagnosis, DSM-5 is lacking cut-off scale, which in turn making diagnosis difficult, especially in young children and early ASD case.¹⁰ The use of DSM-5 is more challenging for diagnosing mild ASD cases, and it have been suggested that there is a decrease in the number of cases when diagnosis is using DSM-5 compared to DSM-4.¹¹ When using the CARS as a diagnostic instrument in a clinically referred sample, a cut-off for ASD is known to improve diagnostic agreement among the CARS and clinical judgment for both 2-year-old and 4-year-old samples. The use of simple instruments will simplify the diagnostic process and may increases the accuracy of the diagnosis.¹²,¹³

The original CARS2-ST is available in English language. Several validated translation of CARS2-ST are already exists in countries such as Japan, India, Brazil and Sweden.⁷,⁸,¹⁴,¹⁵ Studies have
noted that translated version of psychological instruments are being used for various studies worldwide with questionable reliability and validity. The fact that study of construct validity of Indonesian version of CARS2-ST is still scarce made it necessary to investigate further.

METHODS

Study Design
A cross-sectional study was conducted in Child Development Centre in Surabaya, Indonesia from August 1 to December 30, 2019. Total-sampling method was used to enroll participants. Participants inclusion criteria for this study are children aged between 2 and 6 years old with speech and behavior complaints and having parents or guardians who agreed to sign for informed consent. To diagnose ASD, criteria for code 299.0 in Diagnosis Statistical Manual (DSM-5) was used. CARS2-ST protocols were administered twice consecutively by psychologists and pediatricians. Items were scored based on direct observations, caregiver report, and chart review. This study was approved by Ethics Committee Universitas Airlangga Institutional Review Board (170/EC/KEPK/FKUS/2019).

DSM-5 Criteria for Diagnosing ASD
In DSM-5, five criteria are used to diagnose ASD; A) Persistent deficits in social communication and social interaction across multiple contexts, currently or by history, and B) Restricted, repetitive patterns of behavior, interests, or activities, currently or by history. The remaining DSM-5 criteria for diagnosing ASD were outside of CARS2-ST items, which are C) Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities or may be masked by learned strategies in later life). D) Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning. E) Disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay, because the frequent co-occurrence Intellectual disability and ASD; to make comorbid diagnoses of ASD and intellectual disability, social communication should be below that expected for general developmental level.

The Childhood Autism Rating Scale 2- Standard Indonesian Language Version
The Childhood Autism Rating Scale 2- Standard Version (CARS2-ST) is a 15-item observation-based rating scale designed to accurately differentiate children with autism from those with developmental delays without features of autism. Original CARS2-ST is available in English version. To make it available in Indonesian language, the CARS2-ST is translated by experts in Pusat Bahasa Universitas Airlangga. Forward and backward translation is performed to maintain question context. Preliminary pilot test simulation then performed to the translated version to assess the expectable validity and reliability. As a part to assess the overall validity, construct validity is assessed in this study using principal component analysis and varimax rotation to assess whether the translated version have same variable load compared to the original version. All process of translation follows the guideline for psychiatric instruments suggested by Gudmundsson.

Using the instrument, examiners rate the frequency, intensity, duration, and atypicality of the specified behavior while considering the chronological age of the child. Each of the 15 items is rated on a seven-point scale (1, 1.5, 2, 2.5, 3, 3.5 and 4). Higher scale indicates more severe abnormality based on observed children age. A total score is determined by summing the ratings on all 15 items. CARS total scores ranged from as low as 15 (within normal limits on all items) to as high as 60 (severely abnormal on all items). Results are categorized into three category based on score: 1) no ASD symptom (total score < 30), 2) mild-moderate ASD symptoms (total score = 30 - 36.5), and severe symptoms (total score ≥ 37) 6 The usage of the CARS2-ST in this study is authorized by Western Psychological Service (WPS) Rights and Permission with License No. WPS-001447.

Data Analysis
Factor analysis with principal component analysis (PCA) and varimax rotation were performed to identify the number of components of the scale and the variance. Factor analysis were done to assess CARS2-ST items that are hypothetized to fit with DSM-5 ASD criteria. Hence, we hypothesized that translated CARS2-ST items must correspond with the first two dimensions of ASD in DSM-5 (“deficits in social communication and social interaction” and “restrictive repetitive behavior and sensory sensitivities”). Because of its construction in two dimensions, it was expected that ASD could be explained by two factors. In this study, factor loading > 0.4 are considered significant. Higher factor loading are considered to correlate more with corresponding factor.

RESULTS
Two hundred and one children enrolled in the study were those who met the inclusion criteria. There are more male subjects than women, both in subjects whose DSM-5-based examination results are under ASD (77.3%) or not ASD (74.1%). There were no differences in the age range in both ASD (25-72 months) and non-ASD (24-72 months) subjects. Nearly half are first-born (48.5% in ASD children and 48.9% in non-ASD children). Most of them are only children or only have one sibling. Table 1 presents the sample demographic data. Data from 201 respondents were analysed by PCA. We extracted two factors that explained 51.73% of item variance (eigenvalue >1.0). The effect of additional factors was negligible. Table 2 gives the factor loadings of the Varimax-rotated two-factor solution. The responses to items in the
suggested domains are generally appropriately associated with the factors. The two factors explained 44.59% and 7.14% of the item variance respectively. The constructed domains are based on suggested criteria in DSM-5, which matched with that of the underlying factors found by the factor analysis. The first factor can be grouped in “social communication and interaction”, accounted for 44.59% of the variance. The first factor content includes relating to people, imitation, body use, object use, visual response, fear and nervousness, verbal communication, non-verbal communication, and general impressions. The second factor can be grouped in “restrictive repetitive behavior and sensory sensitivities”, accounted for 7.14% of the variance. The second factor consists of emotional responses, adaptation to change, listening response, taste, smell, touch response and use, activity level, and consistency and level of intellectual response. Table 2 presents the principal component analysis result with varimax rotation of factors extracted according to the Kaiser Normalization.

We found that two of loading factors which hypothesized to best fit in “restrictive repetitive behavior and sensory sensitivities” factor were found to be fit better in the “social communication interaction” factor. These loading factors are “general impressions.” and “visual responses”.

Table 1: Characteristic of the sample

| Variable                                      | ASD (+)* n = 66 | ASD (-)* n = 135 |
|-----------------------------------------------|-----------------|------------------|
| Gender, n (%)                                 |                 |                  |
| Male                                          | 51 (77.27)      | 100 (4.07)       |
| Female                                        | 15 (22.73)      | 35 (25.93)       |
| Age (months), median (min-max)                | 56 (25-72)      | 51 (24-72)       |
| Age of complaints (months), median (min-max)  | 24 (12-60)      | 26 (10-72)       |
| Birth order, n (%)                            |                 |                  |
| First                                         | 32 (48.48)      | 66 (48.88)       |
| Second                                        | 19 (29.78)      | 45 (33.33)       |
| Third                                         | 15 (22.74)      | 24 (17.79)       |
| Number of siblings, n (%)                     |                 |                  |
| None                                          | 27 (40.90)      | 56 (41.48)       |
| One                                           | 22 (33.33)      | 53 (39.25)       |
| Two                                           | 14 (21.21)      | 20 (14.81)       |
| Three and more                                | 3 (4.6)         | 6 (4.46)         |
| CARS2-ST Score and Interpretation             |                 |                  |
| < 30 (None-minimal ASD symptom)               | 9 (4.48)        | 133 (66.17)      |
| 30 - 36.5 (Mild-moderate ASD symptoms)        | 44 (21.89)      | 2 (0.99)         |
| ≥ 37 (Severe ASD symptoms)                    | 13 (6.47)       | 0                |

*Diagnosis is based on DSM-5 criteria for ASD

DISCUSSION

Appearance of two unidimensional factors which correspond to the assignments of CARS2-ST items to our hypothesized domains is considered as evidence for the construct validity of the Indonesian version of CARS2-ST. Prior to this study, factor analysis has been done for the original, English version of CARS2-ST. Based on recent report, in the original study it was found that a two-factor solution best fits the loading factors. The study identified two factors that were highly similar to those identified here (“social communication and interaction” and “restrictive and repetitive behavior”), with some differences in specific item loadings. Another study also with similar methodology also found two-factor solutions. The study identifies two domain that hypothesized to correlate with the two factors, named “social communication and sensory issues” and “emotional issues”. Other factor analysis using Promax (oblique) rotation based on DSM-4 showed three-factor methods instead, which divided into “social communication”, “emotional reactivity”, and “stereotyped behaviors and sensory sensitivities”. This showed that, based on DSM of any version, the original version and Indonesian version of CARS2-ST are able to help diagnose stereotyped behaviors and sensory sensitivities.
Construct validity assessment is crucial for a psychological instruments because it tests whether a psychological instruments relate to measures of other constructs as specified by theory. Due to this reason, it is important that translated version of a psychological instruments must have construct validity as similar as possible with their updated original version. Although full publications are scarce, study showed that factor analysis has also been performed in translated versions of CARS2-ST. In Turkish version, a one-factor solution is obtained by comparing the scale with Autism Behavior Checklist and Clinical Global Impression-Severity of Illness. In the Indian version, five-factor solution were obtained by comparing the scale with the International Classification of Disease, Tenth edition (ICD-10), Binet Kamat Scale of Intelligence - an Indian adaptation of the Stanford-Binet Scale of Intelligence; and Gesell’s Developmental Schedule. Here is obvious that currently available factor solution of CARS2-ST is bound to the specifically-compared ASD diagnostic instruments, suggesting that validated local versions of CARS2-ST still comply with ASD basic diagnostic theories. However, the lack of comparison similar to development of original version suggest that construct validity of translated CARS2-ST is still questionable, although their internal validity is reported to be satisfying.

In this study, we found that at least four aspects correspond to both factors. Two of them, “general impression” and “visual response”, are first originally hypothesized to be best explained as observable signs for “restrictive repetitive behavior and sensory sensitivities”. The result showed that both loading factors are better fit for “social communication interaction”. This deviation from our hypothesis can be explained through possibility that at observer’s perspective, both items are often seen as individual stereotypic behavior rather than communication quality. Some clinicians may rate this item based on the quality of a child’s eye contact. In contrast, others may rate it based on a stereotyped behavior or sensory sensitivity (i.e., atypical visual sensory seeking). In fact, in the original CARS2-ST, “visual response” is included in social communication and interaction factors. Data on recent study showed that visual response and nonverbal communication factors showed that both loading factors are better fit for “social communication interaction”.

| CARS2-ST Item                          | Two-Factor Solution |
|----------------------------------------|---------------------|
| Imitation                              | 0.82                |
| Relating to people                     | 0.77                |
| Nonverbal communication                | 0.69                |
| General impression                     | 0.67 0.47           |
| Objects use                            | 0.67 0.42           |
| Verbal communication                   | 0.66                |
| Visual response                        | 0.62 0.44           |
| Fear or nervousness                    | 0.57                |
| Body use                               | 0.51 0.48           |
| Activity level                         | 0.74                |
| Taste, smell, touch response and use   | 0.62                |
| Emotional response                     | 0.62                |
| Listening response                     | 0.57                |
| Level and consistency of intellectual response | 0.50 |
| Adaptation to change                   | 0.42                |

* Extraction Method: Principal Component Analysis. Rotation method: Varimax with Kaiser normalization, only factor loadings ≥ 0.4 that are reported. Two hypothesized DSM-5 domains matched with the underlying factors found by the factor analysis. Value in bold represents CARS2-ST item with two possible factor solution; highest value is selected for solution grouping.

Our study also faced several limitations during its conduction. First, although we are able to observe more than a couple hundred of children, the prevalence of children with ASD is still less than half compared to non-ASD children. This is the problem with short period cross-sectional study design, hence the need to do this study in longer period of time. Secondly, while this study successfully explained the construct validity of Indonesian version of CARS2-ST through a two-factor solution based on DSM-5 criteria, this study result still can not justify the severity of ASD based on DSM-5 criteria. This means we still can not conclude whether ASD severity is actually tied to worse Indonesian version of CARS2-ST result. Further study is needed to clarify this correlation. Finally, while this study is able to show that CARS2-ST items are able to reflect the first two criteria for ASD based on DSM-5, it still can not be used as a basis to correspond with the remaining three of DSM-5 criteria for ASD. This is mainly due to the lack of comparison method for assessing child intelligence, validating child developmental history, and eliminating possible global or focal
developmental disorder, which must be satisfied to diagnose ASD according to DSM-5 criteria. Hence, making Indonesian version of CARS2-ST currently should not be used alone as a modality to diagnose ASD.

CONCLUSION

We confirmed the construct validity of Indonesian version of CARS2-ST, as shown by correspondence of items that reflects ASD symptoms with the first two criteria of ASD in DSM-5. This study supports the continued use relevance of the CARS2-ST for childhood ASD as supplementary diagnostic tool, accompanying the DSM-5 criteria.

Conflict of interest
The authors declare no potential conflict of interest.

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