

EARLY SCREENING FOR AUTISM SPECTRUM DISORDER IN SERBIA: A PILOT STUDY OF SCREENING INSTRUMENTS FOR PARENTS AND CHILD CARE WORKERS

Duško Stupar¹
Aneta Lakić¹
Jasna Jančić¹
Tijana Antin-Pavlović²
Jasmina Marković³
Miroslava Joksimović³
Dubravka Kobac³
Tanja Matic³
Petar Cvetković³
Lidija Hutović³
Nataša Srećković⁴
Dejan Stevanović^{1*}

UDK: 616.89-008.434.5-07

- 1 Clinic for Neurology and Psychiatry for Children and Youth, Belgrade, Serbia
- 2 Child Psychiatry Office "Dr Selaković", Belgrade, Serbia
- 3 Day care center „Čika Jova Zmaj“, Voždovac, Belgrade, Serbia
- 4 Society for mental health improvement, Belgrade, Serbia

Summary

Introduction: Early diagnosis of autism spectrum disorder (ASD) can lead to early interventions, which may improve developmental and academic outcomes in children with ASD. Early screening is thus of significant importance.

Aims: This study had two aims. First, it was aimed to translate into Serbian five screening instruments for ASD: Modified Checklist for Autism in Toddlers – Revised, (M-CHAT-R), Quantitative Checklist for Autism in Toddlers (Q-CHAT), Infant–Toddler Checklist (ITC), and Early Screening Autistic Traits Questionnaire (ESAT) for parents and the Checklist for Early Signs of Developmental Disorders (CESDD) for child care workers. Second, it was aimed to test the feasibility of the included data from parents and child care workers in early ASD screening.

Methods: The translation and cultural adaptation process included standardized forward and backward translation with pilot testing. For screening within a childcare setting, one day-care center was selected. Data from at least one parent and a child care worker were collected for 47 children aged 28.94 months on average (SD = 8.39).

Results: The face and content validity of each version of the instruments is satisfactory. The correlations among the scores showed that the ESET, M-CHAT-R, Q-CHAT, and ITC evaluate slightly different aspects of ASD. The M-CHAT covers similar aspects to the CESDD. Eight (17%) children were positively screened for ASD with at least one instrument. All positively screened with the ESAT, Q-CHAT, or ITC were also positive with the CESDD, while not all positively screened with M-CHAT-R were detected with the CESDD. All five instruments were able to detect the child with confirmed ASD in the final stage – clinical assessment.

Conclusions: All five screening instruments are targeting ASD symptoms at early stage of life in our population. It is feasible to include reports from parents and child care workers in early ASD screening and there is an added value of combining data from the two.

Key words: toddlers, autism, screening, instruments, daycare.

INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by persistent deficits in social communication and social interaction across multiple contexts, accompanied by restricted, repetitive patterns of behavior, interests, or activities [1]. The hallmark symptoms of ASD are deficits in social-emotional reciprocity, deficits in nonverbal communicative behaviors, and deficits in developing, maintaining, and understanding relationships, with restricted, repetitive patterns of behavior, interests, or activities, with or without accompanying intellectual impairment and language impairment. Severity levels for ASD could range from “requiring support” to “requiring very substantial support” [1]. However, developmental, academic, and socioeconomic outcomes of ASD could vary substantially [1, 2].

It is now widely accepted that early diagnosis of ASD can lead to early intervention, which may improve developmental and academic outcomes in children with ASD [2, 3]. Early diagnosis implies that a child with ASD is detected in earlier years of life, even during the first, and he/she is placed on an appropriate intervention. In this regard, screening for ASD is of significant importance and past research showed that detecting children at risk for ASD is possible through the use of screening instruments at early stages of life [4,5]. For this purpose, various instruments were developed [4,6,7], which measures to various degrees of developmentally relevant traits and behaviors related to ASD, including joint attention, pretend play, social communication, stereotyped behaviors, sensory interests, and language development.

In Serbia, there is no screening instrument developed for children at the earliest age. The Children’s Communication Checklist-2(CCC-2), which may be used for ASD screening, is only available in Serbian, but for children aged 4 years and above [8]. This study had two aims. First, it was aimed to translate and culturally adapt into Serbian five screening instruments for ASD: Modified Checklist for Autism in Toddlers – Revised, (M-CHAT-R) [9], Quantitative Checklist for Autism in Toddlers (Q-CHAT) [10], Infant–Toddler Checklist (ITC) [11], Early Screening Autistic Traits Questionnaire (ESAT) [12], and Checklist for Early Signs of Developmental Disorders (CESDD) [13]. Second, it was aimed to test the feasibility of including data from parents and child care workers in early ASD screening.

METHODS

The study had two phases: translation and cultural adaptation and screening within a childcare setting. The Ethics Committee of Clinic for Neurology and Psychiatry for Children and Youth Belgrade approved the study.

Instruments

Modified Checklist for Autism in Toddlers – Revised (M-CHAT-R). The M-CHAT-R is the first part of a two-step ASD screener with 20 items scored as Yes/No [9]. The total score of ≥ 2 positive items might indicate a child at high risk of developing ASD, although there are several scoring options. For the child with ≥ 2 score, it is suggested to complete the second part, the follow-up scale.

Quantitative Checklist for Autism in Toddlers (Q-CHAT). The Q-CHAT is an

ASD screener with 25 items scored using a 5-point scale of frequency, with scores ranging from 0 to 4 [10]. Half the items were reverse-scored. The scores from all items are summed to obtain a total Q-CHAT score, higher scores indicative of more autistic traits. There are some data indicating that the Q-CHAT total score of ≥ 37 is indicative that that child is at high risk of developing ASD.

Infant-Toddler Checklist (ITC). The ITC is an ASD screener with 24 items with three to five choices about developmental milestones of social communication. The ITC is a standardized tool that, in addition to screening cutoffs, has standard scores at monthly intervals for from 6 to 24 months [11].

Early Screening Autistic Traits Questionnaire (ESAT). The ESAT is an ASD screener with 14 items are scored as Yes/No [12]. Children with negative answers on at least 3 of the 14 items be screen-positive and thus at high risk of developing ASD.

Checklist for Early Signs of Developmental Disorders (CESDD). The CESDD is an ASD screener intended for use by child care workers [13]. It has 25 core, plus 4 items. A preliminary cut-off score for ASD was set on 2 or more for children younger than 12 months and 4 or more items for children older than 12 months.

Translation and cultural adaptation. All instruments were translated and culturally adapted for the Serbian language following the same procedures in order to develop versions equivalent to the originals but culturally sensitive, too. Specifically, the instruments and permissions for use were obtained from the developers and copyright holders. The team for translation consisted of researchers familiar with psychological constructs and translators. Two members independently translated the instruments from English into

Serbian. From these two versions, a single form of each instrument was developed (Reconciliation I), which was then translated back into English by two independent English-Serbian translators. A single back translation was developed for every instrument (Reconciliation II) that was compared with the originals (Harmonization) by the principal author and two members not previously included. The entire process resulted in the pre-final versions that were pretested in semi-structured interviews with a group of 7 parents of children with ASD. Afterwards, an expert panel met to evaluate the content and face validity of the versions, the results of the pretesting, and the equivalence with the originals.

Procedures

For screening within a childcare setting, a one day-care center was selected. It was decided to use reports from parents and child care workers who regularly care for children aged 12–48 months. Several meetings were held with parents, child care workers, and language/speech/occupational therapists prior to the testing in order to inform all parties about the screening. Only children with parental consent obtained were included.

Parents completed the M-CHAT-R, ESAT, ITC, and Q-CHAT, while child care workers completed only the CESDD. The parents completed their set of the instruments at home. For all child care workers, a short educational course about ASD was first organized and how to complete the CESDD was instructed. It was arranged that all the children positively screened by at least one instrument should be included in a detailed psychiatric assessment.

RESULTS

For the Serbian versions of the instruments, all the items were felt to be comprehensive, precise, and relevant for assessing ASD in young children, so they were unchanged and no items were added, replaced or omitted in the versions developed.

In the second phase, out of 70 families invited to participate, data from at least one parent and a child care worker were collected for 47 children (67.1% response rate). The children were aged 28.94 months on the average (SD = 8.39, 53.2% boys). Total scores for all instruments and given in Table 1.

Except for the M-CHAT-R Total and CESDD Total ($r = 0.73$), all correlations between the scores were low to moderate, although statistically significant ($p < 0.05$; Table 2). Negative correlations with the ITC Total were due to different scaling options.

Considering cut-off values for the instruments, 8 (17%) children were positively screened for ASD with at least one instrument. The percentages of positively screened children with each instrument are provided in Table 3.

The absolute agreement in screening between parents and child care workers based on cut-off values was assessed by the intraclass correlation coefficient (ICC; Table 4). It was observed that the CESDD

Table 1 Total scores for all instruments

Instrument/score	N	Min	Max	M	SD
CESDD Total	47	0	23	1.45	4.17
ESET Total	47	0	3	0.57	0.68
M-CHAT-R Total	47	0	11	1.06	1.82
Q-CHAT Total	47	10	58	25.57	9.22
ITC Total	46	37	57	50.43	5.23
Social composite	46	13	26	22.09	3.07
Speech composite	46	5	14	12.24	2.46
Symbolic composite	46	12	19	16.11	1.46

Table 2 Pearson's correlations among the scores

Instrument	1	2	3	4	5
1. CESDD Total	/				
2. ESET Total	0.39	/			
3. M-CHAT-R Total	0.73	0.46	/		
4. Q-CHAT Total	0.53	0.29	0.59	/	
5. ITC Total	-0.61	-0.29	-0.52	-0.38	/

Table 3 Positive screening for ASD

Instrument	N (%)
CESDD Total	5 (10.6)
ESET Total	1 (3.1)
M-CHAT-R Total	6 (12.8)
Q-CHAT Total	2 (4.3)
ITC Total	3 (6.5)

and ITC have the highest absolute agreement (ICC = 0.73) for positive ASD screening, which implies that a majority of positively screened with the CESDD would be rather detected with the ITC than with the ESET, M-CHAT-R, or Q-CHAT.

All 8 children positively screened with at least one instrument were sent for a detailed clinical assessment. However, there was a report provided for only one male child in whom ASD was confirmed. All five instruments had the total score positive for ASD screening in that case. For all other children, parents rejected to take part in the final step, the clinical assessments, and provide us with information whether a clinical assessment was performed elsewhere, even though they had agreed to have a follow-up of their children.

DISCUSSION

In this study, five ASD screening instruments were translated and culturally adapted for early ASD screening among Serbian children.

The results of the pretesting phase showed that the face and content validity of each version of the instruments is satisfactory and that all the items are targeting ASD symptoms at an early stage of life in our population. However, ana-

lyzing the correlations among the scores it is indicative that the ESET, M-CHAT-R, Q-CHAT, and ITC evaluate slightly different aspects of ASD, what was also observed before [14,15]. The M-CHAT-R covers similar aspects to the CESDD.

Further analyses indicated that different numbers of children were found positive for ASD with the instruments. Eight (17%) children were positively screened for ASD with at least one instrument. All positively screened with the ESAT, Q-CHAT, or ITC were also positive with the CESDD, while not all positively screened with M-CHAT-R were detected with the CESDD. The CESDD and ITC had the highest absolute agreement in detecting positively screened individuals and all five instruments were able to detect the child with confirmed ASD in the final stage. Taken together, these findings might indicate that each of the instruments have false positive results, as already reported [14–16], and no instrument shows satisfying power in discriminating ASD from non-ASD. However, when combining the CESDD and ITC chances are possibly minimized that false positive children would be selected. On the other hand, it is also possible that when combined the M-CHAT-R and the CESDD more cases would be detected. Nevertheless, these and similar

Table 4 Absolute agreement in screening between parents and child care workers

	ICC*
CESDD vs. ESET	0.31
CESDD vs. M-CHAT-R	0.49
CESDD vs. Q-CHAT	0.55
CESDD vs. ITC	0.73

* *Intraclass correlation coefficient*

assumptions should be further tested when specificity and sensitivity was developed for each instrument.

Finally, the present study showed that it is feasible to include reports from parents and child care workers when early screening for ASD and there is an added value of combining reports from the two [13]. However, there are several challenges that should be considered. First, the response rates could be low for screening, thus additional measures should be taken to include more parents. The same applies for the steps after the screening, because the greatest majority of parents of the children with positive ASD screening failed to participate or

provide information about detailed clinical assessments. Third, there is a high risk of false positive results, thus combining two or more instruments or using follow-up measures would be necessary.

Future work should consider testing the psychometric properties of the instruments, especially specificity and sensitivity, in larger samples of young children, considering different settings (e.g., pediatricians) and including different groups of children (e.g., age groups or children at high risk for ASD). In addition, specific age and gender norms for the Serbian versions should be developed in order to minimize false results of early ASD screening.

RANI SKRINING AUTISTIČNOG SPEKTRA POREMEĆAJA U SRBIJI: PILOT STUDIJA SKRINING INSTRUMENTATA ZA RODITELJE I VASPITAČE

Duško Stupar¹

Aneta Lakić¹

Jasna Jančić¹

Tijana Antin-Pavlović²

Jasmina Marković³

Miroslava Joksimović³

Dubravka Kobac³

Tanja Matić³

Petar Cvetković³

Lidija Hutović³

Nataša Srećković⁴

Dejan Stevanović^{1*}

1 Klinika za neurologiju i psihijatriju za decu i mlade, Beograd, Srbija

2 Ambulanta za dečiju psihijatriju "Dr Selaković", Beograd, Srbija

3 Vrtić „Čika Jova Zmaj“, Voždovac, Beograd, Srbija

4 Udruženje za napredak mentalnog zdravlja, Beograd, Srbija

Kratak sadržaj

Uvod: Rana dijagnoza poremećaja autističnog spektra obezbeđuje započinjanje ranih intervencija kojim se mogu unaprediti razvoj i akademska postignuća ove dece. U tom smislu je rani skrining od velikog značaja.

Cilj: Ova studija imala je dva cilja. Prvi cilj bio je da se prevedu na srpski jezik pet skrining upitnika za poremećaj autističnog spektra i to: Modifikovani test za autizam kod male dece – Revidirana verzija, (M-CHAT-R), Kvantitativni test za autizam kod male dece (Q-CHAT), Test za malu decu (ITC) i Upitnik ranog skrininga na autistične osobine (ESAT) za roditelje i Lista ranih znakova razvojnih poremećaja (CESDD) za vaspitače. Drugi cilj bio je da se testira izvodljivost ranog skrininga upotrebom podataka od vaspitača koji brinu u vrtićima o maloj deci (jaslena grupa).

Metode: Proces prevoda i kulturološke adaptacije uključivao je standardizovan prevod sa Engleskog na Srpski i obrnuto, uz pilot testiranje svih verzija. Jedan vrtić je bio uključen u process skrininga. Podaci od najmanje jednog roditelja i vaspitača su prikupljeni za 47 deteta uzrasta 28.94 meseci u proseku (SD = 8.39).

Rezultati: Pregledna i sadržajna validnost svih prevoda upitnika su zadovoljavajuće. Povezanost između skorova pokazala je da ESET, M-CHAT-R, Q-CHAT, i ITC procenjuju delimično različite aspekte poremećaja autističnog spectra. M-CHAT meri slične aspekte kao CESDD. Osam (17%) deteta imalo je pozitivne skorove za poremećaj na najmanje jednom upitniku. Svi koji su imali pozitivne skorove na ESAT, Q-CHAT ili ITC takođe su imali pozitivne skorove i na CESDD, dok nisu svi oni sa pozitivnim skorom na M-CHAT-R imali pozitivne skorove na CESDD. Sva pet upitnika mogla su da detektuju dete koje je imalo potvrđenu dijagnozu poremećaja u poslednjoj fazi - klinička procena.

Zaključci: U našoj populaciji, sva pet upitnika mere simptome poremećaja autističnog spektra u ranoj fazi. Pokazana je visoka izvodljivost ranog skrininga upotrebom podataka od vaspitača koji brinu u vrtićima o maloj deci i postoji dodata vrednost kada se kombinuju podaci od obe grupe.

Ključne reči: mala deca; autizam; skrining; upitnici; vrtić.

References

1. American Psychiatric Association. Diagnostic and statistical manual of mental disorders (DSM-5®). American Psychiatric Pub; 2013.
2. Eldevik S, Hastings RP, Hughes JC, Jahr E, Eikeseth S, Cross S. Meta-analysis of early intensive behavioral intervention for children with autism. *Journal of Clinical Child & Adolescent Psychology*. 2009 May 19;38(3):439-50.
3. Salomone E, Beranová Š, Bonnet-Brilhault F, Lauritsen MB, Budisteanu M, Buitelaar J, Canal-Bedia R, Felhosi G, Fletcher-Watson S, Freitag C, Fuentes J. Use of early intervention for young children with autism spectrum disorder across Europe. *Autism* 2016;20(2):233-49.
4. Zwaigenbaum L, Bauman ML, Stone WL, Yirmiya N, Estes A, Hansen RL, McPartland JC, Natowicz MR, Choueiri R, Fein D, Kasari C. Early identification of autism spectrum disorder: recommendations for practice and research. *Pediatrics*. 2015 Oct 1;136(S1):S10-40.
5. Zwaigenbaum L, Bauman ML, Choueiri R, Kasari C, Carter A, Granpeesheh D, Mailloux Z, Roley SS, Wagner S, Fein D, Pierce K. Early intervention for children with autism spectrum disorder under 3 years of age: recommendations for practice and research. *Pediatrics*. 2015 Oct 1;136(Supplement 1):S60-81.
6. García-Primo P, Hellendoorn A, Charman T, Roeyers H, Dereu M, Roge B, Baduel S, Muratori F, Narzisi A, Van Daalen E, Moilanen I. Screening for autism spectrum disorders: state of the art in Europe. *European child & adolescent psychiatry*. 2014, 1;23(11):1005-21.
7. McConachie H, Parr JR, Glod M, Hanratty J, Livingstone N, Oono IP, et al. Systematic review of tools to measure outcomes for young children with autism spectrum disorder. *Health Technology Assessment*. 2015;19. doi: 10.3310/hta19410.
8. Glumbić N1, Brojčin B. Factor structure of the Serbian version of the Children's Communication Checklist-2. *Res Dev Disabil*. 2012;33(5):1352-9.
9. Robins DL, Fein D, Barton M. Modified checklist for autism in toddlers, revised, with follow-up (M-CHAT-R/F) TM.
10. Allison C, Baron-Cohen S, Wheelwright S, Charman T, Richler J. The Quantitative Checklist for Autism in Toddlers (Q-CHAT): Psychometric Properties. *Journal of Autism and Developmental Disorders*. 2008; 38:1414-1425.
11. Wetherby AM, Allen L, Cleary J, Kublin K, Goldstein H. Validity and reliability of the communication and symbolic behavior scales developmental profile with very young children. *Journal of Speech, Language, and Hearing Research*. 2002 Dec 1;45(6):1202-18.
12. Swinkels SH, Dietz C, van Daalen E, Kerkhof IH, van Engeland H, Buitelaar JK. Screening for autistic spectrum in children aged 14 to 15 months. I: the development of the Early Screening of Autistic Traits Questionnaire (ESAT). *Journal of autism and developmental disorders*. 2006 Aug 1;36(6):723-32.
13. Dereu M, Warreyn P, Raymaekers R, Meirschaut M, Pattyn G, Schietecatte I, Roeyers H. Screening for autism spectrum disorders in Flemish day-care centres with the checklist for early signs of developmental disorders. *Journal of autism and developmental disorders*. 2010 Oct 1;40(10):1247-58.
14. Oosterling IJ, Swinkels SH, van der Gaag RJ, Visser JC, Dietz C, Buitelaar JK. Comparative analysis of three screening instruments for autism spectrum disorder in toddlers at high risk. *Journal of autism and developmental disorders*. 2009 Jun 1;39(6):897-909.

15. Beuker KT, Schjölberg S, Lie KK, Swinkels S, Rommelse NN, Buitelaar JK. ESAT and M-CHAT as screening instruments for autism spectrum disorders at 18 months in the general population: issues of overlap and association with clinical referrals. *European child & adolescent psychiatry*. 2014 Nov 1;23(11):1081-91.
16. Dereu M, Roeyers H, Raymaekers R, Meirsschaut M, Warreyn P. How useful are screening instruments for toddlers to predict outcome at age 4? General development, language skills, and symptom severity in children with a false positive screen for autism spectrum disorder. *European child & adolescent psychiatry*. 2012 Oct 1;21(10):541-51.

Disclosure and contact

Dejan Stevanovic received permissions to use the instruments. None of the authors had conflict of interest.

Dejan Stevanovic,
Clinic for Neurology and Psychiatry
for Children and Youth,
Dr. Subotic 6a, 11000 Belgrade, Serbia,
Email: dejanstevanovic@eunet.rs
Mob: +381 62 55 1979