

ORIGINAL ARTICLE

Screening for autism spectrum disorders in population of young children in Slovakia

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Abstract

OBJECTIVES: The aim of our study was to assess efficacy of using M-CHAT R/F questionnaire for screening for Autism Spectrum Disorders (ASD) in a sample of Slovak population and compare it with objective diagnostic ASD tool ADI-R. M-CHAT R/T comparison with ADOS-2 diagnostic tool will be published and discussed elsewhere.

METHODS: 92 children with ASD, 72 boys (91.2%), and 20 girls (8.9%), age 1.0–6.9 years, who were signed up by their parents for ASD testing at our institution from 2013 to 2016 were enrolled. All participants were screened by M-CHAT questionnaire filled out by a parent and then complex psychological assessment by trained staff was performed. Latter evaluation included ASD diagnostic tools: Autism Diagnostic Observation Schedule – second revision (ADOS-2) and Autism Diagnostic Interview-Revised (ADI-R).

RESULTS: Significant difference of total (sum) score of M-CHAT means between group of children with ASD and without ASD (as diagnosed by using ADOS and ADI-R) was observed ($p < 0.001$). M-CHAT total score significantly correlated with ADI-R A Domain Abnormalities in Reciprocal Social Interaction, ($r = 0.583$; $p < 0.01$); ADI-R B Domain – Qualitative Abnormalities in Communication ($r = 0.357$; $p < 0.01$).

CONCLUSION: We demonstrated that in our sample of Slovak children M-CHAT total score can be used to differentiate between children with and without ASD risk. M-CHAT also well correlated with accepted objective measure for diagnosis of ASD, i.e. ADI-R. Future work is needed to determine the sensitivity and specificity of M-CHAT in screening for ASD in general population of children in Slovakia.

INTRODUCTION

Autism spectrum disorder (ASD) is one of the most common neurodevelopmental disorders in childhood. It is characterized by two core deficits: impairments in social communication and social interaction, and by restrictive and repetitive behavior, activities and interests (American Psychiatric Association 2013).

Prevalence of ASD in the U.S. was estimated to be 1 in 68 children (CDC 2014). U.S. government survey of parents suggests that 1 in 45 children, ages 3 through 17, have ASD (Blackwell 2014). Prevalence of ASD in Slovakia is currently unknown.

Furthermore, since the etiology of ASD is not clear at the present, our ability to prevent the disorder or to treat children with ASD is very limited at this

point. Applied Behavior Analysis is an ASD Evidence-Based therapy that has been used to decrease the problem behavior and to improve skills that are impaired (Virues-Ortega 2010; DeFilippis 2016).

It was observed, that prognosis of patients with this disease depends on early diagnosis and subsequent intervention. Multiple studies have shown that intense therapy provided in the first three and a half years of age seems to yield significant benefit as opposed to later intervention. Zwaigenbaum *et al* (2013) found that the children who received intervention before age 3 had better prognosis than the ones who started after age of 5.

Early diagnosis is crucial for early intervention. First concerns about a child with future ASD diagnosis are noticed by parents typically at 12–24 months of age (Chavarska 2007; Richards *et al* 2016), but median age of ASD diagnosis in the U.S. sample of 1726 children performed at 11 sites monitoring study was found to be at 3.83 years of age (Christensen *et al* 2016). In Slovakia, in our sample of 254 children who signed up for diagnostic evaluation at Academic Research Center for Autism (ACVA) in 2013 and 2014, median age was 4.51 (Hnilicova *et al* 2015).

Probability of detection of ASD in children under three years old during the well-child visit in Pediatric office, without active screening for ASD is low. Gabrielsen *et al* (2015) observed that licensed psychologists with toddler and autism proficiency missed 39% of cases of ASD during brief, 10-minute long, but highly focused observations. Short, non-focused observation in pediatrician's office may fail to provide enough information to trigger a concern for the pediatrician to refer a child to a specialist. Hence, effective screening methods for ASD early diagnosis are needed.

M-CHAT is a screening technique that was validated in the primary care setting for the age group of 16–30

months. It consists of 20 parent filled “Yes” or “No” questions. It is free and easy accessible on the internet. M-CHAT was translated to Slovak and other languages and has been used in many countries, with culturally specific data available as well. The M-CHAT reported sensitivity and specificity of 0.87 and 0.99, respectively, with positive predictive value of 0.80 and negative predictive value 0.99 (Robins 2001). Updated version M-CHAT R/F was validated on the sample of 18 989 toddlers at Pediatric well-child visits in 2 US geographic regions (Chlebowski 2013).

The aim of our study was to assess efficacy of screening for ASD using M-CHAT R/F Questionnaire in a sample of Slovak population and compare it with objective diagnostic ASD tool ADI-R. M-CHAT R/T comparison with ADOS-2 diagnostic tool will be published and discussed elsewhere.

MATERIAL AND METHODS

The study involved 92 children with ASD, of those 72 boys (91.2%), and 20 girls (8.9%). Their age was 1.5–6.9 years; with mean age 4.0 ± 1.3 years (mean \pm standard deviation). Children were signed up by their parents for ASD testing at our institution. Testing was performed between 2013 and 2016.

All participants underwent screening via M-CHAT questionnaire that filled by a parent and subsequent complex psychological assessment by trained staff. Latter evaluation included ASD diagnostic tools: the Autism Diagnostic Observation Schedule – second revision (ADOS-2) (Lord *et al* 2012) and the Autism Diagnostic Interview-Revised (ADI-R) (Lord *et al* 1994). ASD diagnosis was made based on clinical evaluation and the children had to meet the criteria for ASD on both ASD tools.

ADI-R was evaluated in areas of Abnormalities in Reciprocal Social Interaction, Qualitative Abnormalities in Communication, Restricted, Repetitive, and Stereotyped Patterns of Behavior. ADOS-2 was evaluated in domains of Social Affect, and Restricted and Repetitive Behavior scores.

Software SPSS (IBM 2012) was used for the data analysis. The Student t-test was used to calculate mean differences, and Spearman correlation coefficient was calculated to test the correlation between scores. Values with $p < 0.05$ were considered statistically significant.

The study protocol was approved by the Ethics Committee of the Comenius University Faculty of Medicine.

RESULTS

Significant difference of means of total (sum) score of M-CHAT between group of children with ASD and without ASD (mean score = 4.00 and 8.62 respectively), diagnosed by using ADOS and ADI-R, was observed ($p < 0.001$). M-CHAT differentiated well between those with ASD and those without it. Among those without

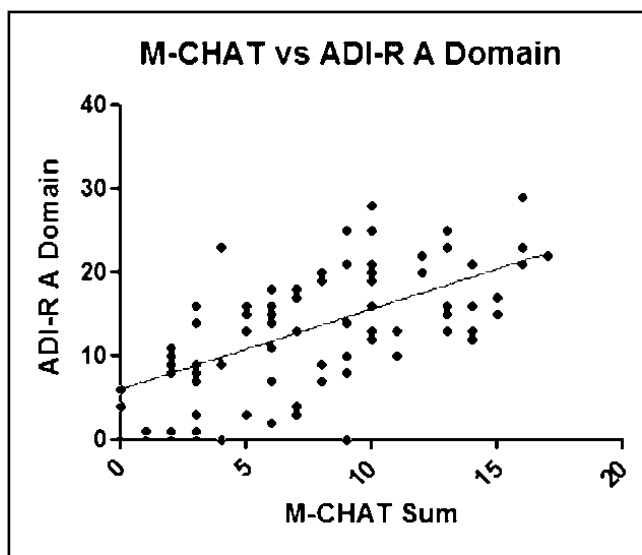


Fig. 1. Correlation between total (sum) score of M-CHAT and ADI-R A Domain in a sample of children with and without autism spectrum disorder ($r=0.583$; $p<0.01$)

ASD, M-CHAT total score 0–5 was found. Medium risk group (ASD, language delay and healthy children), i.e. those with M-CHAT total scores (3–6 points), appropriately pointed to the need for further diagnosis. Total scores which detected high risk group (above 7) were found only in a group of children who were subsequently diagnosed with ASD.

ADI-R – A Domain Abnormalities in Reciprocal Social Interaction, ($r=0.583$; $p<0.01$) which includes failure to use nonverbal behaviors to regulate social interaction, failure to develop peer relationships, lack of shared enjoyment, lack of socioemotional reciprocity significantly correlated with total (sum) MCHAT score (Figure 1).

A significant correlation was found between ADI-R B Domain – Qualitative Abnormalities in Communication ($r=0.357$; $p<0.01$) which includes lack of, or delay in, spoken language and failure to compensate through gesture, lack of varied spontaneous make-believe or social imitative play, relative failure to initiate or sustain conversational interchange and stereotyped, repetitive or idiosyncratic speech (Figure 2).

ADI-R Domain C – Restricted, Repetitive, and Stereotyped Patterns of Behavior containing score of encompassing preoccupation or circumscribed pattern of interest, apparently compulsive adherence to non-functional routines or rituals, stereotyped and repetitive motor mannerisms and preoccupations with part of objects or non-functional elements of material correlated with total MCHAT score ($r=0.357$; $p<0.05$).

DISCUSSION

We herein report first pilot study in Slovakia in which screening for ASD by M-CHAT was compared with objective diagnostic testing for ASD. In our study M-CHAT total score well correlated with deficits in communication and social interaction domains in both objective diagnostic methods, which was similar to previously published results (Albores-Gallo *et al* 2012; Robins *et al* 2014).

In our sample, M-CHAT screening missed 6 (8%) children with ASD; their total score was one point to cut off score. We believe that this is due to limitation of parents-filled screening questionnaires as they are subjective. This number is in accord with previous studies where sensitivity 0.73–0.93 was reported (Garcia-Primo *et al* 2014; Robins *et al* 2014; Nygren *et al* 2012). In our opinion at the time of filling M-CHAT questionnaire, especially if the child is young, parents may not yet be ready to accept that their child differs from other children, so they may deny some symptoms. If the child is their first born, their experience with typically developing children development may be limited.

In the youngest age group, we need to take into account possibility of regress of development. Barger *et al* (2013) in meta-analytic review that included 85 studies ($n=29035$), reported the overall prevalence of

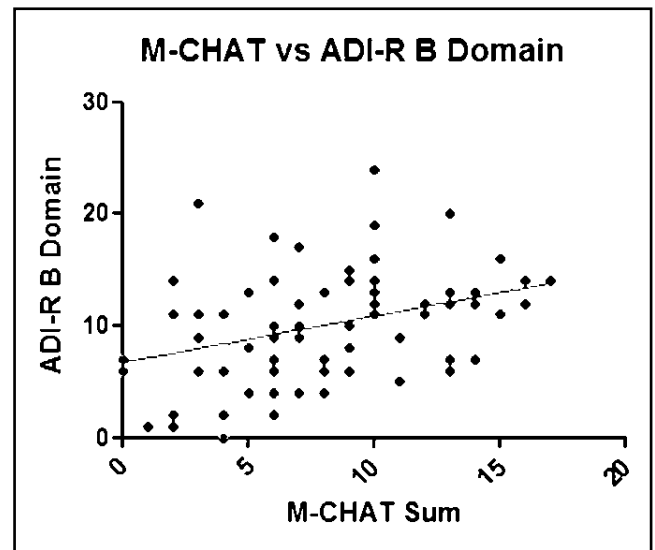


Fig. 2. Correlation between total (sum) score of M-CHAT and ADI-R B Domain in a sample of children with and without autism spectrum disorder ($r=0.357$; $p<0.01$)

regression in ASD to be 32.1% and the mean age of onset of regression as 1.78 years (CI: 1.69–1.89). If the screening questionnaire is filled before this age, it might miss the children who develop ASD in future.

Although it was significant, we observed lower correlation with repetitive behavior in ADI-R when compared to social communication domains. At preschool age, repetitive behavior might be to low degree also present in non-ASD population. Children without autism show atypical behavior at times but in smaller ratio than children who have ASD. (Gabrielsen *et al* 2015). Some components of speech stereotypes and restricted interests may not manifest in such young children especially, in children with language delay. Previously was demonstrated that some of those features cannot be evaluated by objective tools before age of 3 (Lord 1994). Questions about repetitive behavior are very difficult to ask, difficult to explain, and symptoms of repetitive and stereotyped behavior vary a lot among children with ASD, and therefore are often not reported in screening questionnaires.

In the U.S., screening tool M-CHAT-R/F was found to be an effective tool for screening ASD, by reducing the age of diagnosis by 2 years (Robins *et al* 2014). Garcia-Primo *et al* (2014) reviewed use of screening tools in ESSEA Working Groups WG3 titled Testing How Well Screening Instruments Work, in prospectively identifying cases in 20 European countries. M-CHAT was used in 5 countries across Europe as a screening tool of choice, translated and adapted to cultural needs (Brugha *et al* 2011; Campbell *et al* 2017; Canal-Bedia *et al* 2011). In our study in Slovak population sample, the screening M-CHAT showed that it could identify high risk children with ASD. In our opinion it may be

the tool of choice for general population screening for toddlers and preschool children in Pediatric practice.

Although our study is the first in Slovakia to compare screening of children with M-CHAT R/F tool with objective measures, limitations should be acknowledged. First, this pilot study is not a population-based epidemiologic study. The sample was selected based on willingness of parents to participate in ASD diagnostic process at our center. Secondly, the answers to M-CHAT questions might be influenced by the parents as they might already be concerned about child's behavior when they had contacted us or their health professional. Additionally, due to low number of true negative cases, culturally specific sensitivity could not be established.

Despite above mentioned limitations we believe that our results have demonstrated that the M-CHAT summary score can be used to differentiate children with and without ASD and that it correlates in population of Slovak children with generally accepted objective measures for diagnosis of ASD, i.e. with ADI-R. Future work is needed to determine the validity, reliability, sensitivity and specificity of M-CHAT in screening of children for ASD in general population in Slovakia.

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