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Original Article

Accuracy of behavioral responses in early detection of autism spectrum disorders in children aged 18 months to 4 years with speech delay

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Abstract

Background Early detection of autism spectrum disorders (ASD) in children with speech delay is important to improve outcomes. Behavioral responses to calling, teasing, poking, and blocking can be used to screen for ASD in daily practice.

Objective To evaluate the accuracy of behavioral responses to stimuli in detecting ASD in children aged 18 months to 4 years with speech delay.

Methods This cross-sectional study was conducted in children with speech delay aged 18 months to 4 years who visited the Outpatient Clinic of Dr. Cipto Mangunkusumo Hospital, Jakarta. Subjects were stimulated while playing by poking, teasing, calling, and blocking (stopping the child's play using the examiner's hand) and were assessed for their responses. Lack of seeking eye contact with the examiner following the stimulus was considered as a response suggestive of ASD. Independent diagnosis based on DSM-V criteria was considered the gold standard to diagnose ASD or non-ASD.

Results A total of 109 children were included in this study, with an average age of 32 (SD 7.4) months. There were 52 subjects (47.7%) with ASD and 57 subjects (52.2%) with non-ASD. Behavioral response analysis revealed that calling, blocking and teasing had high sensitivity, specificity, positive predictive value (PPV), and negative predictive value (NPV) for identifying ASD. The poking response had the highest specificity to rule out ASD compared to other stimuli, with 75% sensitivity (95%CI 63.2% to 86.7%), 93% specificity (95%CI 86.3% to 99.6%), 90% PPV (95%CI 82% to 99.3%), and 80% NPV (95%CI 70.7% to 89.9%). When all behavioral responses were combined, with lack of a response to all four stimuli considered suggestive of ASD, we obtained 100% specificity, 42% sensitivity, 100% PPV and 65% NPV.

Conclusion The combination of behavioral responses had high specificity, sensitivity, PPV, and NPV for early detection of ASD in children with speech delay. **[Paediatr Indones. 2024;64:17-21; DOI: 10.14238/pi64.1.2024.17-21**].

Keywords: *autism spectrum disorder; children; early detection; speech delay; behavioral response*

Autism spectrum disorder (ASD) is expected to continue to increase in prevalence beyond previous estimates.[1] In 1980, the estimated prevalence of ASD worldwide was 2-4 per 10,000 children; it had increased to 50-60 per 10,000 children in 2010.[2] The most recent United States survey in 2018 revealed an increase in the prevalence of ASD to 16.8 per 1,000 children aged 8 years, the equivalent of 1 in 59 children. Boys are four times more likely to suffer from ASD than girls, with non-Hispanic races suffering more from ASD than Hispanics.[3] The term autism comes from the Greek word meaning "life in one's own world." The term was first introduced by a Swiss psychiatrist, Eugen Bleuler, in 1911, to describe withdrawal due to lack of social interaction in patients with schizophrenia. The definition of autism has grown to include autistic psychopathy, Asperger's syndrome, and infantile autism, which are part of the autism spectrum disorder (ASD).[4]

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ASD-specific screening using the *Modified Checklist for Autism in Toddlers (Revised)* [M-CHAT(-R)] is recommended at 18 and 24 months. A positive screen using the M-CHAT(-R) with Follow-up [M-CHAT(-R)/F] suggests an elevated risk for ASD.[5] Screening for ASD in children younger than 24 months can increase false-positive results compared to screening at more than 24 months of age, but the results can still be informative. Various double-blind randomized studies have shown that early intervention in children under 3 years can improve outcomes, including social attention, intelligence quotient (IQ), language, and severity of symptoms. This shows the importance of early detection in the diagnosis of ASD.[6,7]

Several reports showed that receptive and/or expressive language development abnormality is one of the earliest red flags for ASD.[8,9] The ability to identify this sign in young children is very important, given that the ability to speak before the age of 5 years is a strong predictor of better outcomes in children with ASD. Delayed speaking is often the main motivation of parents to have their children examined.[10] In children with speech delay, a number of behavioral signs can be used to detect early ASD in daily practice, such as the teasing response, calling response, and blocking response.[6,11] Similarly, testing the child's response to poking is commonly used in the observation of a child suspected of ASD. This response is based on hypo- or hyperreactivity to sensory input in children with ASD.[12] The authors theorize that a non-ASD child, when given a poking stimulus, would make eye contact with the person delivering the stimulus or with a caregiver. Such a response would be absent in a child with ASD. As such, we aimed to determine if the response to poking can distinguish ASD from non-ASD in children with speech delay. In addition, we evaluated the sensitivity, specificity, positive predictive value (PPV), and negative predictive value (NPV) of the response to poking in children with ASD compared to other signs, such as responses to calling, blocking, and teasing.

Methods

This cross-sectional study included children aged 18 months to 4 years of age with delayed speech whose parents were willing to participate and provide written informed consent. Children were excluded if they had certain syndromes, congenital abnormalities, chronic diseases (cerebral palsy, general developmental disorders, Down syndrome, hydrocephalus), history of premature birth (less than 37 weeks), or who had received sensory integration therapy for ≥ 1 month or at least 8 meetings.

While playing, subjects underwent stimulation by poking, teasing, calling, and blocking, and their responses were assessed. The poking stimulus was given on the upper right arm from behind without the subject's knowledge, while the subject was playing, with slight pressure of 0.5 to 1 second on the arm. The teasing stimulus was done by giving a toy to the subject, then pulling it back. The calling stimulus is done by calling the subjects name while playing, while the blocking stimulus was done by directly stopping the child's play using the examiner's hand. The expected typical response from the subject to each of these stimuli was to make eye contact with the examiner. An atypical response, i.e., lack of eye contact, was considered suggestive of ASD. Subjects were then grouped into either ASD or non-ASD groups based on DSM-V criteria by two child neurology consultants, independent from the examiner administering the aforementioned stimuli.[13] The study was conducted at the Child Neurology Clinic, Department of Child Health, Cipto Mangunkusumo Hospital, and Anakku Clinic, Jakarta.

Data analysis was done using *SPSS version 25* (*IBM*, Armonk, New York). We calculated the sensitivity, specificity, PPV, and NPV of each of the calling, blocking, teasing, and poking responses, with diagnosis based on the DSM-V criteria as gold standard. We then calculated the sensitivity, specificity, PPV, and NPV of the combination of all four tests, with an atypical response to all tests considered positive for suspected ASD.

Results

A total of 109 children were included in the study, with an average age of 32 (SD 7.4) months. Eighty-three (76.1%) subjects were male. There were 52 subjects (47.7%) with a diagnosis of ASD and 57 subjects (52.2%) with a diagnosis of non-ASD. Non-ASD diagnoses included expressive language

disorders, social communication disorder (13 subjects; 11.9%), articulation disorders, and deafness (**Table 1**).

The diagnostic performance of the behavioral responses to calling, blocking, teasing, and poking to screen for ASD are shown in **Table 2**. The response to poking had the highest specificity of 93% (95%CI 86.3 to 99.6), with a 75% sensitivity (95%CI 63.2 to 86.7), 90% PPV (95%CI 82 to 99.3), and 80% NPV (95%CI 70.7 to 89.9) for detecting ASD. For the combined response, with a lack of response to all four stimuli considered as indicating suspicion of ASD, the specificity was 100% with a sensitivity of 42% (95% CI 28.8 to 55.7%) (**Table 3**).

Table 1. Characteristics of subjects

Characteristics	(N=109)
Mean age (SD), months	32 (7.4)
Age by group, n (%)	
<24 months	12 (11.0)
24 to <36 months	59 (54.1)
36 to <48 months	38 (34.8)
Sex, n (%)	
Male	83 (76.1)
Female	26 (23.8)
ASD, n (%)	52 (47.4)
Non-ASD, n (%)	
Expressive language disorders	57 (52.2)
Communication disorder	41 (37.6)
Articulation disorders	13 (11.9)
Deafness	1 (0.9)

Discussion

Most subjects were aged 24 to <36 months (59; 54.1%), followed by 36 to <48 months (38; 34.8%). Subjects had a mean age of 32 (SD 7.4) months. The large proportion of subjects aged two years shows increasing awareness and sensitivity to the child's developmental disorders on the part of the parents, as well as their effort to bring the child to a medical professional for further evaluation. A study involving 1,420 children with ASD and 2,098 children with intellectual/developmental disabilities, and found that the median age when parents began to realize the existence of a developmental disorder was 2.1 (range 1.9-2.3) years. In addition, the median age when parents first discuss developmental disorders with healthcare providers is 2.3 (range 2.2-2.5) years. Interestingly, the median of age of a child when a parent is first told that their child has ASD is 5.2 (range 4.9-5.5) years.[14] Although early signs of ASD can be recognized by trained professionals before the age of two years, many children with ASD are not diagnosed until school age.[15] Another interesting finding was that the average time between parental awareness of a potential developmental disorder and the first discussion with a health worker is 0.2 (95%CI 0.1 to 0.3) years. In addition, the median time between

Table 2. Diagnostic test results of various behavioral responses in detecting ASD

Variables	ASD (n=52)	Non-ASD (n=57)	Sensitivity (95%CI) %	Specificity (95%CI) %	PPV (95%CI) %	NPV (95%CI) %
Calling response						
(-)	35	8	67 (54.6 to 80)	86 (76.9 to 94.9)	81 (69.7 to 93)	74 (63.6 to 84.7)
(+)	17	49				
Blocking response						
(-)	41	6	78 (67.7 to 89.9)	89 (81.5 to 97.4)	87 (77.6 to 96.7)	82 (72.7 to 91.7)
(+)	11	51				
Teasing response						
(-)	38	9	73 (61 to 85.1)	84 (74.7 to 93.6)	80 (69.6 to 92.1)	77 (67 to 87.8)
(+)	14	48				
Poking response						
(-)	39	4	75 (63.2 to 86.7)	93 (86.3 to 99.6)	90 (82 to 99.3)	80 (70.7 to 89.9)
(+)	13	53				

Table 3. The combined results of the four diagnostic test examinations for detecting ASD

Variables	ASD (n=52)	Non-ASD (n=57)	Sensitivity (95%CI) %	Specificity (95%CI) %	PPV (95%CI) %	NPV (95%CI) %
Test result (-)	22	0	42 (28.8 to 55.7)	100 (100 to 100)	100 (100 to 100)	65 (55.5 to 75.5)
Test result (+)	30	57				

the first discussion with a health professional and the age at which an ASD diagnosis was made is 2.7 (range 2.5-3.0) years.[14]

In our study, there were more male than female subjects. This is consistent with previous research that reported that one of the risk factors for delayed speech is male sex. In addition, other risk factors for delayed speech include family history of delayed speech, prematurity, low birth weight, low parental education level, and large family size.[13] We found that of 109 subjects with delayed speech, 52 (47.7%) had ASD and 57 (52.2%) had non-ASD.

Failure to respond when called upon can be an initial manifestation of a decline in social orientation that is typical in children with ASD. Consistent failures of the child to respond to his name being called from the beginning of life can be part of a further development cascade, which in turn can lead to social communication disorders in children with ASD.[15] Infants who do not respond to his/her name tend to be less involved in social activities from time to time. Additionally, responding when called is included in all diagnostic criteria for ASD.[16] In our study, the response to calling to detect ASD in children with speech delay had 67% sensitivity (95%CI 54.6 to 80), 86% specificity (95%CI 76.9 to 94.9), 81% PPV (95%CI 69.7 to 93%), and 74% NPV (95%CI 63.6 to 84.7%). Similarly, a previous study noted that the response to calling had 70% sensitivity (95%CI 50 to 90), but a lower specificity of 70% (95%CI 62 to 78%).[11]

In their literature review, Martinez et al. stated that the majority of children with ASD and developmental disorders have accompanying stereotypical behaviors occurring at a level that can hinder the development of academic and social behavior. However, the management may be difficult, because this stereotypic behavior is often maintained by automatic reinforcement.[17] Several studies have shown that procedures based on reinforcement, such as differential and non-contingent reinforcement, can reduce stereotypical behaviors.[17] In addition, other study have shown that penalty-based procedures, such as responses when inhibited, can be a necessary component of management.[18] To the best of our knowledge, there have been no studies on the diagnostic value of responses to blocking in detecting ASD. In our study, the response to blocking to detect ASD in children with delayed speech had 78% sensitivity (95%CI 67.7 to 89.9%), 89% specificity (95%CI 81.5 to 97.4%), 87% PPV (95%CI 77.6 to 96.7%) and 82% NPV (95%CI 72.7 to 91.7%).

Teasing requires the ability to understand intentions, non-literal communication, faking, and social context. Children with ASD have difficulty in this regard, and, as a result, they also have difficulty realizing teasing.[19] In our study, the diagnostic test results of the response to teasing to detect ASD in children with speech delay were 73% sensitivity (95%CI 61 to 85.1), 84% specificity (95%CI 74.7 to 93.6), 80% PPV (95%CI 69.6 to 92.1), and 77% NPV (95%CI 67 to 87.8).

We also assessed the response to poking to detect ASD in children with delayed speech. The poking response had 75% sensitivity (95%CI 63.2 to 86.7%), 93% specificity (95%CI 86.3 to 99.6%), 90% PPV (95%CI 82 to 99.3%), and 80% NPV (95%CI 70.7 to 89.9%) for detecting ASD. Compared to the other three responses, the response to poking had the highest specificity (93%) for detecting ASD, with a sensitivity of 75%. One of the common symptoms of children with ASD is a sensory processing disorder, characterized by an excessive or lack of responsiveness to sensory stimuli. Abnormalities in response to sensory stimulation are found in 95% of children with ASD. This may explain the high diagnostic performance of poking, a tactile stimulus.[14]

Interestingly, when the four behavioral response examinations are combined, with an absent response all four of the stimuli considered as indicating suspicion of ASD, a very high specificity (100%) with 42% sensitivity (95%CI 28.8 to 55.7) was obtained. This finding implies that a child who does not respond to all four stimuli can be ascertained to have ASD. Responses to poking, calling, teasing, and blocking can be used in a busy clinical practice for the early detection of ASD in children with delayed speech. With earlier diagnosis, management can be given in a timely manner in order to improve long-term developmental outcomes.

Conflict of interest

None declared.

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