

Diagnostic Accuracy of the Children's Communication Checklist-Persian in Identifying Children with Autism Spectrum Disorder

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Abstract

Background: This study examined the diagnostic accuracy of the children's communication checklist-Persian version (CCC-Persian) in differentiating children with autism spectrum disorders (ASD) from typically developing (TD) children.

Methods: The parents of 47 children with ASD and the parents of 104 TD children completed the CCC-Persian. The children were monolingual Persian-speakers between 5 and 11 years of age. The sensitivity, specificity, likelihood ratios, and cut-off score of the CCC-Persian were calculated in identifying children with ASD.

Results: The mean pragmatic composite score (PCS) of the CCC-Persian was significantly lower in children with ASD than in the TD children ($P>0.05$). Corresponding cut-off score, sensitivity, and specificity were 107, 86%, and 97%, respectively. The positive and negative likelihood ratios were obtained as about 30 and 0.14, respectively.

Conclusion: The CCC-Persian has the potential to be used as a valid clinical tool for diagnosing pragmatic language impairment or screening ASD in Persian-speaking children.

Key Words: Autism, Children's Communication Checklist, Pragmatic deficit, Sensitivity, Screening, Specificity.

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1- INTRODUCTION

Pragmatics refers to the use of language in social interactions (1). Children with Autism Spectrum Disorder (ASD) are deficient in their pragmatic language ability (2), which causes communication problems in these children (2-5).

Looking at pragmatics as a component of social communication, children with ASD suffer from different pragmatic problems (2, 6). Inability to perceive the context of communication, and impaired perception of facial cues or body language, and an inability to infer and predict consequences are considered pragmatic deficits in children with ASD (6). Other pragmatic deficits observed in children with ASD include selecting and maintaining social relations, modifying vocabulary in a conversation, staying on topic, requesting explanations, managing communication styles in discussions, turn-taking, and incorporating the audience's perspectives. Excessive talking, interrupting others, and insensitive social responses are also observed. Consequently, they may experience repeated communication failures in their daily interactions (7-10).

Two types of tools are currently being used for evaluating pragmatics in children (3). The first group refers to the formal tests such as the Comprehensive Assessment of Spoken Language (CASL) (11) and the Test of Pragmatic Language-2 (TOPL-2) (12). While these tests provide valuable information regarding children's pragmatic ability in formal controlled conditions, they fail to consider the extensive and diverse contexts in which the children should communicate in natural ways (3). On the other hand, informal tools such as Pragmatic Protocol (1) systematically observe the children's language use in natural settings. Furthermore, they provide vital information concerning the different pragmatic domains of the children's

behavior in various communication styles (1).

The Children's Communication Checklist (CCC) is a structured informal instrument (13, 14) that can differentiate between 4-7-year-old typically developing (TD) children and those with pragmatic problems in the same age range (13, 15). Also, it has been shown that the CCC can be used to screen children with ASD differentiating them from TD children (16, 17). The CCC has been translated and adapted in different languages, including English (13, 18, 19), Dutch (20, 21), Norwegian (22), Spanish (23), Finnish (24), Thai (25), and Persian (26, 27). Studies have shown that the pragmatic composite score (PCS) of the CCC, which is a combined score of the CCC's pragmatic components, can identify pragmatic deficits in ASD children (19, 20) (**Table 1**). Usually, children with ASD have communication disorders, particularly in pragmatics (19, 20, 25). The PCS can accurately differentiate between children with and without ASD (16, 17, 19).

Pragmatic deficits in children with typical developmental conditions may be independent of their language disorders (22). Children with ASD are diagnosed with explicitly compromised weaknesses in pragmatics (2). According to previous studies, 100% of children with ASD and 10.5% of TD children showed the corresponding clinical symptoms of pragmatic deficits when their parents completed the CCC (19, 28).

Because pragmatic skills are culture-dependent (3), we need culturally adapted tools to assess pragmatics in different societies. A small number of studies exists in this area in Persian (26, 27, 29-34). The majority of these studies have only focused on a limited number of pragmatic skills in children with a certain disorder or specific age range. None of these studies has

investigated the diagnostic accuracy of the tools.

The first edition of the CCC (19) was adapted into Persian in 2007 with internal consistency between 0.75-0.84 in children between 5 and 11 years of age (27). In 2014, an adapted version of the CCC-2 to Persian showed 0.66-0.74 internal consistency in children between 7 and 9 years of age (35). A factor analysis study was performed on a combined list of Persian adapted checklists of the CCC and the CCC-2 in search of the best assignment of items to different subscales. This

process ultimately resulted in the current Persian version of the CCC, called the CCC-Persian (26).

To identify the clinical applicability of the CCC-Persian, the current study followed a three-fold purpose; first to determine the test-retest reliability of the CCC-Persian, second to find out whether the PCS can differentiate children with ASD and TD, and third to define the cut-off scores and diagnostic properties of the PCS in Persian-speaking children between 5 and 11 years of age.

Table-1: A list of diagnostic studies using the CCC for comparing different clinical groups

Study	Diagnosis	n	Range of age (year)	Result of phase I of diagnostic accuracy: Significant difference on PCS	Index test	Reference standard	Phase II of diagnostic accuracy		
							Cut-off	Sensitivity	Specificity
Bishop (19)	ASD from SLD	17	5-17	Yes	-	-	140	-	-
Geurts et al. (20)	ASD from TD	50	5-14	Yes	-	-	123	-	-
Chuthapisith et al. (25)	ASD from TD	50	4-6	Yes	CCC -Thai	Pediatric psychiatrist	132	94%	86%
Charman et al. (17)	ASD from children with special educational needs	119	9-13	Yes	CCC	ADI	132	93%	46%

Note: Phase I of a diagnostic accuracy study = comparison between the population with typical language development vs. those diagnosed with ASD; Phase II = the diagnostic properties of a clinical tool (36). ASD = Autism Spectrum Disorder; TD = Typically Developing children; ADI = Autism Diagnostic Interview.

2- MATERIALS AND METHODS

2-1. Participants

The participants of the study included 147 children in two groups of case and control. Those in the control group (TD group) were recruited from 30 elementary schools and kindergartens that were randomly selected from two urban areas with middle-class socioeconomic status in Isfahan, Iran. The SLP randomly selected

six students from each school, one student from each grade level, following the inclusion procedure explained in 2.1.1. Of the 180 children who were initially referred to the pediatric psychiatrist by the SLP, 151 children were evaluated by the pediatric psychiatrist. Twenty-nine parents were not referred to the pediatric psychiatrist for no reason and were excluded from the study (Figure 1).

All participants in the case group (ASD group) were recruited from five autism rehabilitation centers in Isfahan city by the same SLP. Of 75 children with inclusion criteria who were asked to refer to the pediatric psychiatrist, 62 children were

evaluated by the pediatric psychiatrist. Thirteen parents refused to see the psychiatrist due to cultural beliefs and were excluded from the study. In total, 213 out of 255 invited children (83.5%) participated in the study.

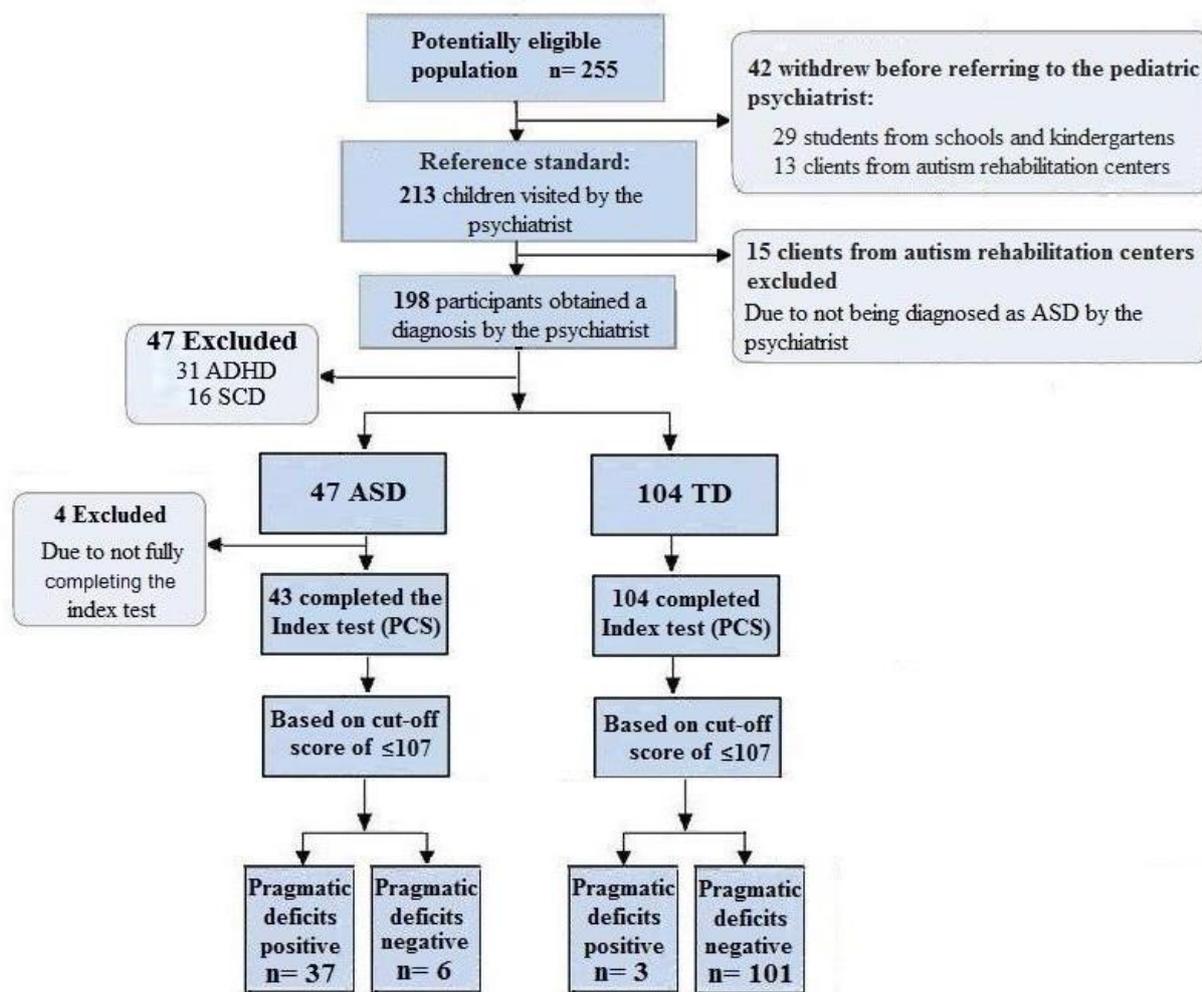


Fig. 1: Participant Flow Chart Following Standards for Reporting Diagnostic Accuracy Studies (STARD)

2-1-1. Diagnostic process

All children were first assessed by an SLP and then referred to a pediatric psychiatrist to receive the diagnosis. The SLP was responsible for the recruitment process and received the informed consent forms. The SLP also monitored the children’s referrals to the pediatric psychiatrist and the completion of the CCC-Persian by their

parents. The whole process of labeling and allocating the participants to the case and control groups was conducted by the pediatric psychiatrist.

2-1.2. Inclusion and exclusion criteria

Monolingual Persian-speaking children with no history of hearing impairment, speech or language disorders, physical, emotional, or intellectual disabilities

(except in the group of children with ASD) were included in this study. In the group of participants with ASD, only children who had verbal communication or good non-verbal communication were included. Therefore, in this study the severity of ASD ranged from mild to moderate. Children with severe ASD were not included in the current study. As for the group of children with ASD, the diagnosis was made based on the DSM-5 criteria. No criteria for gender inclusion were considered. The parents must have been able to read and respond to the questions. The exclusion criteria included not getting diagnosed by the pediatric psychiatrist, having a diagnosis other than ASD, and not responding to more than six items (10%) of the CCC-Persian (37). None of the children with TD had a history of speech therapy or other interventions that affected the outcome. Children with ASD were selected by an expert speech and language pathologist (SLP) and an expert pediatric psychiatrist based on DSM-5 criteria for ASD.

2.2. Reference standard

Recently, different tools have been adapted and developed to screen Persian-speaking children with ASD in Iran including the Gilliam Autism Rating Scale (GARS), Autism Behavior Checklist (ABC), Gilliam Autism Rating Scale (GARS-2), the short version of the Checklist for Autism in Toddlers (Q-CHAT-10), the Hiva scale, and Modified Checklist for Autism in Toddlers (M-CHAT) (38-42). However, these tools have not been widely used among health practitioners to diagnose children with ASD and pragmatic deficits. On the other hand, diagnostic accuracy studies usually employ the best available reference as standard instead of the "gold standard" when there is no universal agreement among the professionals (43). This study used the current standard practice for diagnosing ASD in Iran; and a psychiatrist makes the

diagnosis based on the criteria listed in DSM-5.

2.3. Index test

The current version of the CCC-Persian has 69 items divided into ten subscales, including A) speech, B) syntax, C) inappropriate initiative, D) coherence, E) stereotyped language, F) use of contexts, G) compatibility, H) social interaction, I) interests, and J) non-verbal communication. The items have either positive or negative meaning, and the parents need to select one answer out of four multiple choices, including "0 = cannot judge", "1 = never or rarely", "2 = sometimes", and "3 = always". Then, the sum score of each subscale is subtracted from a base score of 30, making the score of each subscale less or more than 30 according to the strengths or weaknesses of the child's communication skills. The pragmatic composite score (PCS) is the index test in the current study, which is based on the original CCC. The PSC is calculated by summing up the scores of five pragmatic subscales, C to G. The lower the PCS scores, the more extensive the problems are (19, 26, 27). No prior cut-off score was determined since one of the main aims of the study was to investigate the empirical cut-off scores derived from the real data. In fact, in this study, we examined whether the PCS could detect pragmatic deficits in children with ASD.

2.4. Procedure

According to the Declaration of Helsinki (44), all parents signed a written informed consent prior to participation. The same SLP assessed all participants following the procedures explained above and referred them to the pediatric psychiatrist. To identify the test-retest reliability, the parents of 45 children (30%) were randomly selected to re-complete the checklist after a two-week interval. This number was randomly selected from both groups of the participants.

2.5. Data Analysis

The intraclass correlation coefficient, version 3, 1 (ICC_{3,1}) (45), was used to examine the test-retest reliability. The mean of the PCS of the CCC-Persian was calculated based on the Q-Q plot fulfilled criteria of the normal distribution and the difference between the groups was investigated by the use of t-test.

Since the CCC-Persian is a new test, we analyzed some diagnostic accuracy properties, including sensitivity, specificity, positive and negative likelihood ratios, and the area under the curve (AUC), in distinguishing children with ASD from TD ones. To further investigate how well the PCS can distinguish children with ASD from TD, the Receiver Operating Characteristic (ROC) curve analysis was conducted. A ROC curve shows the values of the sensitivity (i.e., the ratio of positives that are correctly diagnosed as having ASD compared to the sum of children with ASD who were correctly diagnosed and children with ASD whom the PCS misdiagnosed) and the specificity (i.e., the ratio of negatives that are correctly diagnosed as not having ASD compared to the sum of children without ASD who were correctly diagnosed and children without ASD whom the PCS misdiagnosed) and the cut-off points of the PCS. The current study calculated likelihood ratios with a confidence interval (CI) of 95% through the sensitivity and specificity results. According to the related literature (46), positive likelihood ratios of 10 or more and negative likelihood ratios of less than 0.1 are considered large and definite and are very informative. A positive likelihood ratio of 5 to 10 and a negative likelihood ratio between 0.1 and 0.2 are moderately informative. A positive likelihood ratio between 2 and 5 and a negative likelihood ratio between 0.2 and 0.5 are relatively informative. If a positive likelihood ratio falls below 5 or a negative ratio is over

0.5, the test results will not be informative (46). In this study, the AUC was also calculated, which is a scale for measuring the overall correct diagnostic power. The interpretation of the AUC results is that AUC values between 0.9-1 are considered as excellent, values between 0.8-0.9 as good, values between 0.7-0.8 as fair, values between 0.6-0.7 as poor, and AUC values between 0.5-0.6 as failed (47).

3- RESULTS

3-1. Patient characteristics

Two hundred and thirteen children (97 girls, age range: 5-11 years) were candidates of participation in the present study, and the data of 147 finally included children (73 boys and 74 girls) were analyzed. With a consideration of the STAndards for the Reporting of Diagnostic accuracy studies (STARD) recommendation for reporting diagnostic studies (48), the flow diagram of participants is shown in **Figure 1** indicating that 78%, and 2.9% of children with ASD, and TD have pragmatic deficits, respectively. The demographic data of the groups and the mean of PCSs have been shown in **Table 2**.

3-2. Pragmatic outcomes

The results of the t-test show that the two groups significantly differed in terms of PCS (**Table 2**). In fact, children with ASD scored significantly lower than the TD children ($P < 0.001$). Also, the effect size based on Cohen's *d* shows that the size of mean differences between TD children and children with ASD is large (49). The two groups were not significantly different in terms of age ($P > 0.05$).

3.3. Diagnostic accuracy of the CCC-Persian

To investigate the diagnostic accuracy of the CCC-Persian, the AUC value of PCS of the CCC-Persian was created with a corresponding cut-off score for the ASD group as 0.945 (95% CI: 0.89-0.99), which

indicates its overall accuracy in differentiating the children with ASD from the TD group (**Table 3** and **Fig. 2**). As for the PCS diagnostic values, the cut-off score of the PCS, sensitivity, specificity, LR+, and LR- obtained to distinguish ASD children from TD children were 107, 86%, 97%, 29.65, and 0.144, respectively (**Table 3**).

In other words, distinguishing children with ASD from TD, LR+, and LR- were

very informative, and also the AUC value was strong, indicating that the odds of a child's pragmatic competence being affected by ASD is about 30 times more than a typically developed peer. In fact, the PCS can differentiate children with ASD from TD children with great power. Also, the results of the test-retest reliability by using ICC_{3,1} ranged from 0.82 to 0.96 showed that all subscales of the CCC-Persian had an ICC higher than 0.75.

Table-2: Comparison of the Pragmatic Composite Score (PCS) as a clinical marker of pragmatic deficit across the two groups

Clinical groups	Age M ± SD	PCS (M ± SD)	95% CI	Mean Difference	Cohen's d	p
Typically Developing (n=104, 37 boys)	7.89 ± 1.91	121.06 ± 6.29	[119.84, 122.29]	23.8*	2.55	p < 0.001
Autism Spectrum Disorder (n=43, 36 boys)	7.75 ± 1.84	97.2 ± 11.59	[93.64, 100.77]			

Note. PCS = Pragmatic Composite Score; CI = Confidence Interval; * The mean difference is significant at the .05 level. ** Cohen's *d* of .1, .25, and .4 representing small, medium, and large effect sizes, respectively (49).

Table-3: The diagnostic features of Children's Communication Checklist-Persian for children with ASD compared to TD children

Diagnosis of ASD (n = 43) from	AUC	Cut-off of PCS	Sensitivity (95% CI)	Specificity (95% CI)	LR+ (95% CI)	LR- (95% CI)
TD (n = 104)	0.945	≤107	86 % (0.72-0.93)	97 % (0.91-0.99)	29.65 (9.7-91.5)	0.144 (0.06-0.30)

Note. ASD = Autism Spectrum Disorder; TD = Typically Developing; AUC = Area Under the receiver operating Curve; CI = Confidence Interval; PCS = Pragmatic Composite Score; LR+ = Positive Likelihood Ratio; LR- = Negative Likelihood Ratios.

4- DISCUSSION

This study analyzed the reliability and diagnostic accuracy of the CCC-Persian. It also determined the cut-off score of a composite score of pragmatic skills in Persian-speaking children with ASD and TD ones from 5 to 11 years of age. The results showed that the checklist has excellent reliability and contains outstanding diagnostic features that can differentiate among children with and without pragmatic deficits and ASD. The

current study can confirm the logicity of using the CCC-Persian by the Persian speech-language pathologists in clinical decision-making for children with pragmatic problems, particularly children with ASD. The effect size suggests that the PCS can differentiate between children with ASD and TD ones with a large statistical power. The results showed that 78% of children with ASD have pragmatic deficits, while Bishop and Baird reported that 100% of ASD children have pragmatic

deficits (19). This relatively large difference is probably due to the differences in ASD definitions and the diagnostic tools. Bishop and Baird used the Autism Diagnostic Interview-Revised

(ADI-R) to diagnose ASD (19), but in the present study, the clinical judgment of a pediatric psychiatrist (based on DSM-5) was used for diagnosing children with ASD.

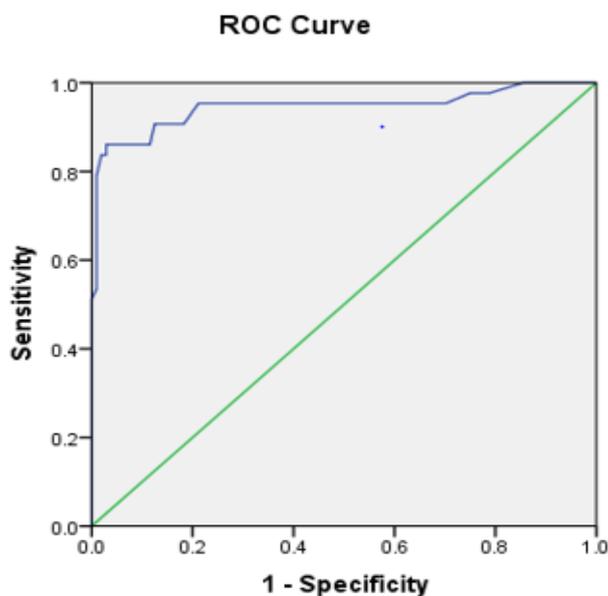


Fig. 2: ROC (Receiver Operating Characteristics) curve analysis of the Pragmatic Composite Score (PCS) for differentiating the pragmatic deficit in children with ASD from TD children

Our study and that of Bishop & Baird (19) have shown that pragmatic problems are very common in children with ASD. However, the content of screening checklists in Iran often deals with stereotyped, repetitive, and limited behaviors and interests, and less attention is paid to the pragmatic problems of these children (38-42). The CCC-Persian, however, can identify pragmatic problems resulting from ASD in Persian-speaking children, and in screening ASD, it pays close attention to the inclusion of pragmatic problems.

Our findings demonstrated that the CCC-Persian has a good diagnostic validity. The original English CCC introduced a cut-off score of 132 for distinguishing the children with SLI from those with pragmatic language impairment (13). The Thai version of the CCC also obtained the same score for distinguishing the pragmatic

problems in children with ASD from the typically developing peers (25). In the present study, a cut-off score of 107 was obtained to distinguish children with ASD from TD children, which is lower than those reported in the other studies regarding the original and Thai versions (20, 25). This large difference is most likely due to a structural change in the CCC-Persian; because, as mentioned before, the present checklist is the result of a factor analysis on a combination of items from the original version and the second version of the CCC (26). Therefore, the number of subscales, as well as the number of items of some sub-scales, have changed compared to the original CCC (19, 26). Obviously, the range of PCS changes in line with the changes in the number of items in the subscales, and thus the results of the CCC-Persian are not comparable to the original CCC.

The corresponding diagnostic features of the reported cut-off score, 107, including the sensitivity (86%) and specificity (97%) with an overall accuracy measured by AUC as 0.94 which is high enough to be considered excellent and acceptable, were similar to the Thai version of the CCC (25). As the range of AUC varies from zero to one, closer values to one represent a better overall accuracy of the test in identifying the target group (50). The sensitivity and specificity in the Thai version of the CCC were 94% and 86%, respectively (25). In another study, the AUC, the sensitivity, and the specificity of the CCC in English were reported 0.79, 93%, and 46%, respectively (17). In addition to the structural differences between the CCC-Persian and the original CCC, it is argued that the different diagnostic tools might be responsible for the difference between the cut-off scores in different languages. For example, Charman et al. used the ADI-R and ICD-10 (17), and Chuthapisith et al. applied the assessment of two developmental and behavioral pediatricians to diagnose ASD (25). In the current study, we used a pediatric psychiatrist who diagnosed children with ASD. Considering the results, the LR+ more than 10 in our study ensures the clinicians with the high precision decision that the PCS equal to or lower than 107 comes from a child with ASD by about 30 times more than a TD child. The LR- of less than 0.2 suggests how often it is likely that a typically developing child has a pragmatic deficit. The LR- of less than 0.1 is the most suitable of this assumption (51). So far, no study has reported the LRs of the first version of the CCC, and the results of this study are not comparable to studies using the second version of the CCC (CCC-2), since the compositions of the subsets of CCC-2 and CCC-Persian are different.

Bishop and Baird showed that with increasing the age of children, the PCS shows a significant improvement (19). In

the present study, the mean age of the participants in the two groups was not significantly different. So, the effect of age on the PCS was controlled, and the difference in the PCS of the two groups can be attributed to the children's communication performance.

The clinical application of the CCC-Persian for screening children with ASD is supported by the robust diagnostic features, i.e., AUC and likelihood ratios. The lower score means that the child is more likely affected by a pragmatic deficit caused by ASD or even affected by ASD. The findings of the present study showed that pragmatic deficits are common in children with ASD, which is in line with the results of previous studies (2, 19). So, it can be interpreted that if pragmatic deficits are identified in a child through the CCC-Persian, we should also consider the probability that this child may have such a disorder. The sensitivity and specificity of the cut-off score obtained in this study are strong. So, if a child's PCS is equal to or less than 107, the SLP should be suspected of ASD, and it is reasonable to refer the child to a pediatric psychiatrist for further evaluation. So, similar to Charman et al. (17) and Deckers et al. (16), we argue that the PCS can be used as a screening tool for ASD. Unlike other screening checklists available for screening ASD in Iran, the CCC-Persian emphasizes pragmatic problems.

In sum, the CCC-Persian has the potential to be used as a clinical tool to assess pragmatic language impairments in Persian-speaking children with or without ASD. Also, based on the results of this study, the PCS of the CCC-Persian can accurately differentiate between children with and without ASD, who demonstrate signs of pragmatic problems. In general, the findings are consistent with other studies on the first version of the CCC (17, 19). We recommend that children who are

underscored be referred to a pediatric psychiatrist for a final diagnosis.

4-1. Limitations of the study

Our study was limited regarding the evaluation of children by one pediatric psychiatrist. We encourage researchers to use a second diagnostic opinion for future studies. Contrary to the parents of children with ASD, many parents in TD groups were reluctant to visit the specialist due to a sense of criticism or having denial regarding their children's condition, leaving us with missing participants in this group. We could not replace them due to the lack of time and funding. Furthermore, it is suggested to conduct further studies on different severity levels of ASD with larger sample sizes. Comparing teachers' viewpoints with parents provides detailed information about the agreement between two of the most influential parties in children's lives. So, it is suggested that this area be considered in future studies. Another limitations of the present study was that the effect of participants' IQs was not considered; though, according to Chuthapisith et al. who did not find any correlation between IQ score and the CCC score in ASD groups (25), it may be concluded that the results of the present study are not questioned in this regard.

5- CONCLUSIONS

Like many other languages, a limited number of tools are used in Persian to evaluate pragmatic abilities in children. This study provides empirical evidence for the clinical use of the CCC-Persian for spotting pragmatic difficulties among Persian-speaking children. The CCC-Persian is an easily-administered and structured informal assessment tool that employs the parents' observation to report children's communication behaviors in natural environments. The CCC-Persian was confirmed to have a reliable diagnostic competence that is reflected in its strong psychometric measures. Also,

we suggest that the CCC-Persian has the potential to be used as a first-level screening tool for identifying children with ASD whose prominent signs are within the pragmatic domain. They are candidates for an in-depth further diagnostic assessment. Thus, the results of this study indicate that children with ASD can be distinguished from TD children with quite a high precision by the CCC-Persian. Unlike other screening checklists available for screening ASD, the CCC-Persian emphasizes pragmatic problems.

6- ACKNOWLEDGMENTS

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7- ETHICAL CONSIDERATIONS

This study is part of a large study that has been approved by code 1396.3.250 in the local Institutional Review Board of the Isfahan University of Medical Sciences. All procedures performed in studies involving human participants were under the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards which were approved by the ethics committee of Isfahan University of Medical Sciences. According to the Declaration of Helsinki (44), all parents signed a written informed consent prior to participation.

8- FUNDING

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9- CONFLICT OF INTEREST

None.

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