A Methodology for Exploring Experiences of Individuals with Tourette Syndrome in Oral and Silent Reading Assessments

Aileen C. Bautista*, **Macario O. Cordel II, Cely D. Magpantay,**
Catherine Manuela L. Ramos
De La Salle University, Manila, Philippines
aileen.bautista@dlsu.edu.ph*

Abstract

Tourette Syndrome (TS) is a neurological condition characterized by tics, and extant literature reports that it has an impact on the cognitive and linguistic performance of the learners. However, limited research has examined the oral and silent reading experiences of individuals with TS. Our study explored the experiences of individuals with TS and the typically-developed cohort (TD) when taking oral and silent reading assessments. Utilizing the Neale Analysis of Reading Ability - II (NARA- II) to compare the TS and the TD cohort, the data demonstrates similar performance in most areas for the TS cohort. Nevertheless, the data reveals that the TS group experiences more difficulty in taking an oral reading comprehension test compared to silent reading. Thus, the study is significant in that it sheds light on the experiences of the learners in a reading comprehension assessment that may provide pedagogical assistance to this group of learners. The proposed methodology serves to advance our understanding of spoken language processing in individuals with Tourette Syndrome.

1 Introduction

Tourette Syndrome (TS) is a neurodevelopmental disorder that results in tics (American Psychiatric Association | American Psychiatric Association. DSM-5 Task Force, 2013) characterized by involuntary and repetitive, motor movements, or oral vocalizations (Jung and Révész, 2018; American Psychiatric Association | American Psychiatric Association. DSM-5 Task Force, 2013). Typically, TS manifests during childhood, and its severity would peak during adolescence and sometimes wanes during early adulthood (Greene et al., 2016). TS affects the prefrontal cortex and basal ganglia (Thibault et al., 2008) which potentially impacts individuals' memories (Clark and Lum, 2017; Takács et al., 2018) and indirect language processing (Legg et al., 2005). In the Philippines, TS appears to be considered a non-prevalent movement disorder, affecting 0.9% of TS patients(Yu et al., 2021). Studying the potential learning challenges experienced by individuals with TS is critical as they remain an integral part of the community and thus, deserve attention and support. Despite this, to our knowledge, no studies have been conducted to investigate the specific impact of TS on reading comprehension. Thus, this paper aims to describe the oral and silent reading performances of individuals with TS.

The subsequent sections of this study encompass a comprehensive analysis of various aspects related to reading comprehension and TS. Firstly, the "Reading and Noticing" section reviews the intricate relationship between TS and the cognitive processes involved in reading, with a particular focus on attentional mechanisms and the ability to notice and comprehend textual information. Secondly, the "Working Memory" section explores the role of working memory in reading performance for individuals with TS, examining the cognitive process and overall comprehension. Next, the "Data Gathering and Experiments" presents the research methodology, including participant selection, data collection techniques, and experimental procedures employed in the study. Finally the "Results and Analysis," which presents the empirical findings obtained from the study, analyzing the data collected to uncover insights into the relationship between TS and reading comprehension.

2 Related Works

2.1 Reading and Noticing

Reading is a vital skill that should be developed among learners, even second language (L2) learners. As a result, various L2 reading activities are aimed at helping learners achieve development in comprehension ability and L2 acquisition (Jung and Révész, 2018). One important strategy in L2 reading, as supported in different studies, is the

glossing of words which is related to noticing.

In addition, various investigations have explored how text processing takes place among learners and how it is related to noticing, which is associated with the glossing of words during reading (Zuo, 2021).

Interestingly, Truscott (1998) describes noticing as a consciousness-raising process deemed necessary for learning. Capitalizing on the learners' consciousness, awareness, or attention results in being input-focused (Truscott, 1998; Robinson, 1995). Thus, it allows learners to absorb the input being looked at, which is deemed a requirement for language learning. However, extant debates on the effectiveness of noticing were raised by some scholars, as language acquisition is asserted to be an unconscious process, which was first posited by Krashen (1985). Furthermore, a few argue that, despite being a requirement for successful learning, noticing does not guarantee linguistic form and structure acquisition, particularly the syntactic and semantic aspects (Truscott, 1998; Lee, 2021). Even though it intensifies awareness of a given task, it does not automatically suggest conscious attention to the details of what should be learned (Schmidt, 1990). However, extant literature suggests that noticing is more effective in phonetic and orthographic forms of a text, primarily when the learners are not guided and are left to read the material independently.

Schmidt (1990) asserted that by noticing, learners become aware and focused on the linguistic stimuli, which may yield an understanding of the text. Hence, noticing is the building block for reading comprehension, an important language skill. This essential claim is supported by the arguments of Logan et al. (Logan et al., 1996) that "people will learn about the things they attend to, and they will not learn much about the things they do not attend to" (p. 620).

Corollary to this, De Vos et al.(De Vos et al., 2019) explains that from the various scholarly explorations on noticing, it can also be in the form of "noticing a gap", which means that once the learners realize that the input is missing from their linguistic inventory, especially in the aspect of vocabulary, all the more attention is paid to that input takes place; thus, remembering or retention may take place. Through noticing, strengthening the vocabulary bank may be possible.

2.2 The Working Memory

The prefrontal cortex and the basal ganglia region store these cognitive inputs in short-term working memory (Grossberg, 2018), while the eyes prolong their attention on a set of words through noticing. Thus, it may potentially be stored in long-term memory. Robinson (1995) proposes that memory regulates what can and cannot be successfully acquired and learned. Hence, every learner is wired to acquire linguistic inputs from the act of noticing. But, Robinson (1995) argued that memory regulates what can be successfully acquired and learned and, likewise, what inputs the brain cannot retain. Therefore, if there is an aberration in the part of the brain that oversees the memory systems, it can be gleaned that individuals may suffer from language learning challenges. In particular, individuals with short-term memory may be affected by their neurological conditions, including those with TS.

Several studies, e.g. (Eddy et al., 2012; De Nil et al., 2005; Legg et al., 2005) have been conducted to investigate the working memory of people with TS, particularly their cognitive and language abilities. Eddy et al. (2012) reported that both adults and children with TS display cognitive challenges in the areas of executive function and working memory, as evidenced by attention deficit and verbal fluency. In their investigation, they concluded that learners with TS showed deficiencies in their verbal fluency and working memory. Furthermore, compared to the non-TS cohort, the said group produced fewer words.

Meanwhile, in the research of Debes et al. (2011) that used Weschler Intelligence scales among children with TS, they reported that the TS cohort has a "lower IQ score" (p. 146) and a lower Working Memory Index than the typically-developed cohort. Likewise, a stable upward trend was observed among the typically developed group compared to the TS group which exhibited quite an erratic trend. They attributed these to the tics, comorbidity, and/or severity of tics of the said cohort.

For De Nil et al. (2005), who investigated the speech disfluencies of individuals with TS, the results show that both cohorts believed that they had speech disfluencies. For oral reading, both cohorts displayed disfluencies, but the difference did not present any statistical significance. However, the children with TS exhibited more of these issues than the adults. Thus, the older the person with TS,

the fewer disfluencies were observed, aside from their own perceived speech issues becoming fewer as the participants became older.

In the investigation by Legg et al. (2005), they used a language test battery that included The Test of Language Competence and the Fable Interpretation Test. The results show that the TS group displayed difficulty performing language tasks that required higher order thinking; thus, showing that the said group possesses "disturbance" (p. 15) in the prefrontal cortex.

It is evident that there are very few attempts to explore the linguistic capacity of individuals with TS and the challenges they experience in performing such tasks. We believe that delving into such an investigation helps the teachers and the people who aim to help them in their studies and in other cognitive tasks. Thus, this current investigation endeavors to describe the oral and silent reading performances of individuals with TS. In particular, it aims to 1) compare the oral reading performance of learners with TS to that of a typical cohort in terms of reading comprehension, reading accuracy, reading time, and reading rate. 2) identify the oral and silent reading experiences.

3 Data Gathering and Experiments

The methodology is summarized in Fig. 1. This study used the Neale Analysis of Reading Ability - II (NARA-II) (Neale et al., 1999) – a standardized test assessing the various aspects of reading abilities. NARA-II has been widely used as a psychometrically sound reading assessment to provide baseline reading ability for children and adolescents. Meanwhile, Adaptive Behavioral Assessment-3 (ABAS-3) is an instrument that measures adaptive skills across the lifespan. Yale Global Tic Severity Scale (YGTSS) is used to measure the total motor and phonic tics of the individuals with TS.

During the initial stage of the study, an alternative assessment instrument was under consideration. However, during the pre-assessment pilot testing, participants expressed difficulty in comprehending the selected instrument. Consequently, a decision was made to employ the NARA-II as an alternative choice, given its alignment with the reading proficiency of the respondents. Moreover, considering the existing literature highlighting cognitive challenges, especially in higher order thinking, among individuals with TS (Legg et al., 2005), the NARA-II emerged as a suitable standardized

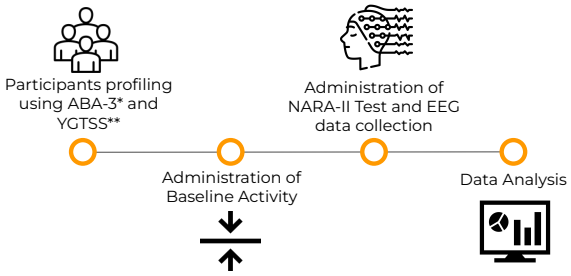

Figure 1: Our proposed methodology includes (1) participants profiling through ABAS-3 and YGTSS, (2) conducting baseline activity, (3) collecting EEG signals while performing tasks in NARRA-II, and (4) data analysis.

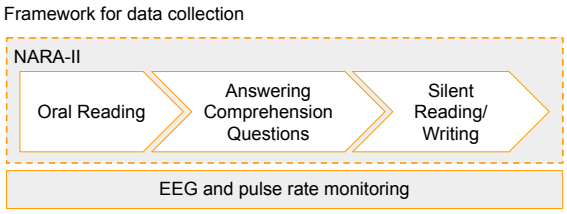

Figure 2: Our data collection setup performs simultaneous EEG monitoring and administration of NARA-II test which include Oral Reading Task, Answering Comprehension Questions and Silent Reading/Writing.

tool for evaluating the multifaceted aspects of reading in this context.

That is, despite the NARA-II being primarily designed for children, preliminary results indicate its suitability for adult participants whose second language is English. These individuals demonstrated similar challenges as expected during different sections of the test, particularly in the oral reading component.

As displayed in Fig. 2, the NARA-II test involved the answering of the two reading areas: 1) oral reading and 2) silent reading. After oral reading, the participants answered a series of oral reading tests. Meanwhile, the silent reading was followed by the writing of the details read from the story that was read silently. Biosignals were collected through EEG and HRV while the reading tests were being administered.

To ensure that the audio and video recordings were collected, the data gathering was aided by a few technological gadgets. First, Audacity was used to record the audio using the built-in microphone of the laptop. Second, using a USB microphone and Logitech c90 ProHD web camera, another video was recorded through OBS studio version 27.2.4.

3.1 Participants

The participants of the study comprised two cohorts. The first group consisted of individuals with TS (N=5) with a mean age of 22. All participants with TS were invited based on the endorsement of a local Tourette Syndrome association, and they were formally diagnosed with TS by their neurologists. Also, the Yale Tic Severity Scale (YGTSS) was used to measure the tics manifesting in each participant. The second group consisted of typically-developed (TD) group (N=5)with a mean age of 25.4. Hence, they are deemed typical developed community samples. Age matching was done to ensure that the participants were of similar age. The TD cohort, on the other hand, has one female member, whereas the TS cohort is entirely male.

For the TS cohort, the following were the screening requirements: a formal diagnosis of TS, no neuropsychiatric disorder, no serious hearing problem, and no serious speech problem. For the TD cohort, the following were the screening criteria: no neuropsychiatric disorder or comorbidity, no serious hearing problem, no serious speech problem, and having a matched age with the members of the TS cohort.

3.2 Experiments

The computation as reflected in the NARA-II manual was used. In particular, the raw score of the following was computed: 1) accuracy; 2) comprehension; and 3) rate. For rate, the following were identified: 1) the number of words read; 2) the cumulated number of words; and 3) the total time in seconds. In addition, the error count was also calculated by listening to the audio recording to identify the following errors: 1) mispronunciation, 2) substitution, 3) refusals, and 4) addition. For silent reading and writing, the details were identified from the answer and their sum was computed.

Two raters were asked to look at the audio and video recordings of the utterances to make sure that any mistakes were found. Then, they listened closely to the utterance to identify the errors and their types. The results were compared, and the recording was listened to in order to identify the actual error if there were disagreements in the types of errors. The accuracy, comprehension, number of words read rate, and reading time rate were compared with each other using the correlation coefficient and its corresponding significance.

Table 1: Average Scores from NARA

Metrics	TS	TD
Accuracy	46.83	59.83
Comprehension	13.00	18.17
Rate		
Words Read	215.83	270.50
Cumulative No. of Words	215.83	146.33
Total Time in Seconds	221.17	270.50
Words per min	64.29	113.35
Total Errors	28.33	38.67

4 Results and Analysis

This section reports the results of this investigation. It is organized as follows: 1) a description of the oral reading performance of the two cohorts in the following areas: reading comprehension, reading accuracy, reading time, and reading rate; and 2) a description of the silent reading performance.

4.1 Oral Reading

As presented in Table 1, the results showed a difference between the two groups in how well they understood what they read. The TS group obtained an average of 46.83, while the TD group obtained an average of 59.83. Interestingly, although the participants in both groups were of different ages, they had trouble remembering specific details from the passages. It was displayed regarding the oral reading rate (average oral reading rate: 64.29, 113.35). These results back up what Takács et al. (2018) said about how people with TD did better than those with TS when it came to how well they remembered words. Furthermore, only one person in the TS group said that his tics and how well he understood what people were saying helped him perform, whereas the other five said that their performance was due to how strong their tics were. However, it appears that it is not statistically supported in our findings.

Further, all respondents admitted that their focus during the oral reading test was the phonological accuracy of the words read during the oral reading instead of the details found in the story. As a result, they had difficulty recalling the information included in the stories. Ergo, the concept of phonological decay is experienced by both cohorts. Despite that, the results show that the TS group experiences faster phonological decay, as reflected in the scores for accuracy. This finding appears to challenge the study of Takács et al. (2018), as

Table 2: The p-values of the reading variables from the TS participants (top) and Non TS participants (bottom). For TS participants, the correlation between comprehension and words read is statistically significant ($p < 0.05$). For Non-TS participants, these variables with strong correlation also include accuracy and comprehension ($p < 0.01$).

	Accuracy	**Comprehension**	**Words Read**	**Reading Time**	**WRpm**
Accuracy	0.0000	0.1072	0.1310	0.6271	0.4478
Comprehension	0.1072	0.0000	0.0155	0.0930	0.8434
Words Read	0.1310	0.0155*	0.0000	0.3200	0.7443
Reading Time	0.6271	0.0930	0.3200	0.0000	0.0992
WRpm	0.4478	0.8434	0.7443	0.0992	0.0000

	Accuracy	**Comprehension**	**Words Read**	**Reading Time**	**WRpm**
Accuracy	0.0000	0.0007**	0.0038	0.4394	0.0743
Comprehension	0.0007	0.0000	0.0110	0.5213	0.0612
Words Read	0.0038	0.0110	0.0000	0.1792	0.2450
Reading Time	0.4394	0.5213	0.1792	0.0000	0.5892
WRpm	0.0743	0.0612	0.2450	0.5892	0.0000

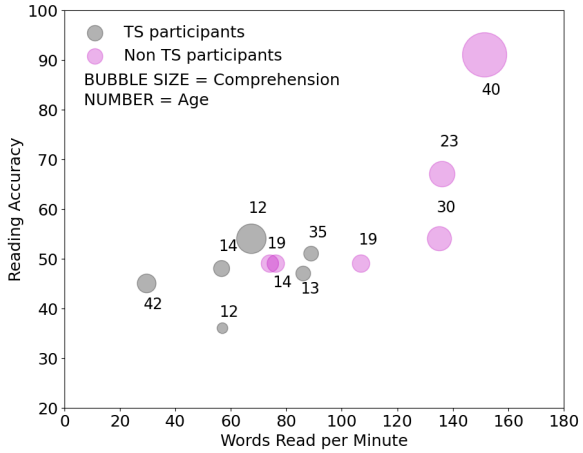

Figure 3: Reading accuracy of TS participants and TD participants as a function of words read per minute. Note that the reading accuracy of the TD participants in our reading activity show a steeper, direct relationship with their reading speed.

they argued that the tics, which are a form of "brain abnormality" (p. 84), cause the enhanced function of the procedural memory, where a possible heightened grammar processing is at play. However, it is significant to emphasize that the participants in their study listened to the words in sequence only, whereas the participants in our study read the words aloud. Therefore, it is possible that the different skills used resulted in polarized findings.

The same thing was true for the comprehension score: the group with TS did not perform better than the group without TS. However, there is only a five-point difference in the average comprehension scores compared to the high difference in the previously mentioned areas. These numbers show that, even though there are problems with the accuracy of the information given, the TS group is almost as good as the TD group when it comes to understanding what is being said in a reading passage orally, thus, accuracy. It supports the past research that, in the area of linguistic accuracy, both cohorts performed almost the same (Takács et al., 2018).

Even though the TS group consistently obtained lower scores for phonological errors, they also had a lower average (28.33) than the TD group (38.67). Both groups displayed more errors in substitutions (87.06% and 81.90%). The TS cohort showed fewer substitution errors (148) than the TD cohort (190). Based on this, it may be safe to conclude that the tics may not seriously affect the utterance, as errors such as substitution, mispronouncing words, omissions, and reversals are also observed even by individuals without TS. Hence, the tics do not hinder individuals with TS from achieving phonological accuracy.

In addition, age as a significant area revealed interesting patterns. On the one hand, our data shows older TD participants have higher words read per minute (WRpM). As shown in Fig. 3, a higher WRpM is approximately exponentially related to reading accuracy and comprehension. It supports the observations made in the study by Cain and Oakhill in 2010, which suggests a possible relationship between linguistic accuracy and performance. However, their study used children as participants. On the other hand, for the TS participants' data, age

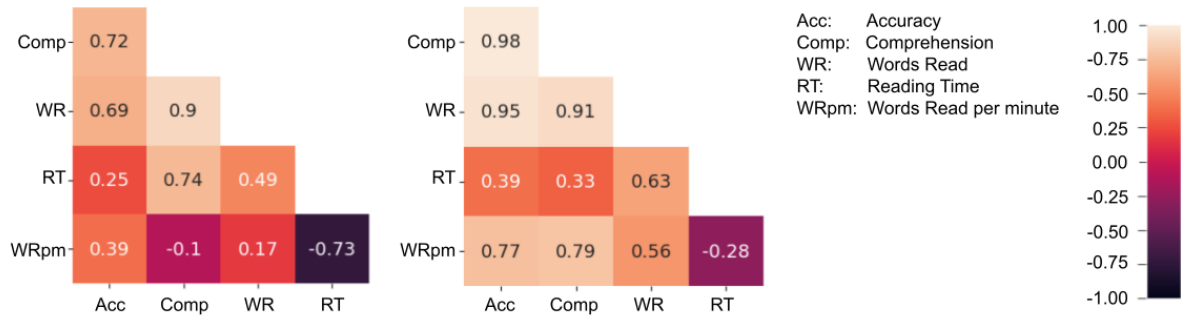

Figure 4: The correlation matrix of the reading variables from the TS participants (left) and TD participants (right). In our study, strong correlations are defined by the absolute value of the correlation coefficient greater than 0.9.

does not provide a clear relationship with WRpM. WRpM shows a weak linear relationship with reading accuracy. However, this relationship is not reflected in the samples' reading comprehension results. Although the number of cases and, thus, the data presented are small, the observation may provide a snapshot of TS cases in the Philippines. The observations here are similar to those discussed in the reading ability study of adults with TS conducted by De Nil et al. 2005 and the investigation of Singer et al. 1995 among children with TS.

For the TD participants, WordsRead, Comprehension, and Accuracy are strongly correlated. As shown in Fig. 4, a fairly strong correlation between WRpM, Comprehension, and Accuracy is also seen. Meanwhile, for the TS participants, only the WordsRead and Comprehension are strongly correlated.

Furthermore, looking closely at the components of NARA, Table 2 shows that the correlation between comprehension and words read for TS and non-TS (TD) participants is statistically significant. However, unlike TS participants, TD participants correlate accuracy and comprehension with $p < 0.01$. This may suggest that for the TS cohort, accuracy and comprehension may not reflect a strong statistical correlation; ergo, it cannot be argued that the said group performed the same compared to the TD.

In Table 3, the Mean SDNN which is the average of the standard deviation of the duration of one heart beat (N-N) or interbeat intervals (IBI) measured in milliseconds (ms) were obtained for each group and each task using the Student's t-test. The Mean SDNN were statistically significant for the TS and TD groups during the oral reading and answering comprehension questions.

The SDNN, which is sometimes referred to as heart rate variability (HRV), was obtained for each participant from the pulse oximeter readings. These readings were measured over the duration of the three different tasks (Oral Reading, Answering of Comprehension Questions and Silent Reading). Participants varied in accomplishing the three tasks. Participants took from 2 to about 10 minutes to finish each task. For the table above, 5 participants from the TS cohort and their counterparts in the TD cohort were included. One pair was excluded because of technical difficulties in recording the audio during the administration of the NARA-II test.

Table 3: Statistical analysis of heart rate variability of TS and TD cohorts under oral reading, silent reading and silent writing. SDNN means Standard Deviation of duration of one heart beat (N-N). OR means oral reading, ACQ means Answering Comprehension Questions, SR means silent reading.

Case	Tasks	Mean SDNN	p-value	95% CI	
				min	max
TS	OR	184	0.0053	64.39	304.36
TS	ACQ	114	0.0146	26.34	203.20
TS	SR	199	0.1728	-135.07	535.05
TD	OR	44	0.0066	14.36	75.16
TD	ACQ	42	0.0037	16.07	69.74
TD	SR	44	0.0813	-8.85	98.28

The comparison of the mean SDNN among the TS and TD groups shows that the TS had a normal heart rate variability with a mean of 144 to 199 ms than the TD group with a lower mean of 42 to 44 ms. Given the present data, the TS cohort is more relaxed in executing oral and silent reading during the experiment. At the same time, the TD is more anxious. Gauging this difference reflect that the TS, regardless of their tics, are more confident in handling oral and silent reading while the TD are

more conscious of their performance. The TS are more prone to anxiety performance than the HRV measures.

4.2 Silent Reading

The self-report data reveals that the participants found the silent reading easier than the oral reading. They all agreed that since the silent reading part excluded oral utterances of the words, they encountered no problems in reading the text. Likewise, most participants retained almost all significant parts of the story, except for one participant who remembered a few words from the story that was read. Unfortunately, this participant failed to rewrite the story in sentence form. When asked after the test, all participants reported that there felt no tics during the silent reading and writing. The findings support the claims of the previous study of Legg et al.2005 that tics may manifest during more difficult parts of the tests that include higher-order critical thinking skills. Since the silent reading activity was measured by story rewriting and this is not considered a critical thinking question, it may be possible that this is the reason why the participants experienced no tics during this test part.

For this part of the test, the TD cohort reported no challenges. They all mentioned that the silent reading activity was easier than the oral reading activity because they also become conscious of their pronunciations during this test. Therefore, the issue of being attuned to linguistic utterances is observed by both cohorts. This suggests that it is natural among test takers to pay particular attention to how they say the words when reading them aloud, and it is not only the individuals with TS who do this because they have tics.

5 Conclusion and Future Work

Our proposed methodology used NARA-II as an instrument and biosignals to investigate individuals with TS during the oral and silent reading tests. The methodology presented allows the quantitative comparison of the reading performance of TD and TS participants. EEG signals, when collected during the reading tasks could facilitate association of TS tics characteristics and the reading performance of TS participants. Using the proposed methodology, preliminary results show that the difference between TS and TD could be numerically described. Some initial results support existing studies, and some reveal polarized outcomes, which motivate further study.

The current investigation possesses a few limitations. First, the sample size of the TS population. Based on the report by Yu et al. (2021), TS is not a common disorder in the Philippines which contribute to the limited number in TS who were also diagnosed properly at an early age. As for the diagnosis, the lack of protocol and proper diagnosis at an earlier age reduced the TS cohort and the variability and demographic characteristics like TS comorbidity. Furthermore, the lack of epidemiological studies on neurodevelopmental disorders in the country, especially TS studies, contributed to the small sample size. Second, the majority of the common literature in TS studies in other countries is related to neurocognitive disorders and comorbidity, but it does not investigate the TS reading component, such as language, and its implications for their learning difficulties. Third, the time frame of the data collection was during the COVID-19 pandemic. Hence, fewer people were willing to participate in a study and attend a face to face assessment.

Ethics Statement

Research ethics clearance were secured prior to the data gathering. Prospective participants about the risks and benefits of the study, along with the procedure of the tasks and what they would do. The participants signed the informed consent form. For adolescent participants who have not reached the age of 18, the consent of the parents was sought.

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