Differences in Pragmatic Language Measures in Mothers of Children with Autism or Fragile X Syndrome

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Mastery of pragmatics, which refers to the language used in different social interactions, is key to effective communication in daily life. However, studies have found that mothers of children with an autism spectrum disorder (ASD) or Fragile X Syndrome (FXS) are at an increased risk for impaired pragmatic language skills. Though many measures of pragmatic language exist for children, very few are available for these mothers. Two commonly used measures for the mothers include the Pragmatic Rating Scale (PRS) and the Pragmatic Language Subscale of the Broad Autism Phenotype Questionnaire (BAPQ). This study, unlike previous ones that only look at one measure, compares these two measures using cutoff scores. Subjects who score above a certain value are classified as "positive" for pragmatic language issues, and agreement refers to when the subject is classified positive by both measures. While high agreement was expected given the common purpose of the two measures, the results showed very low agreement. This was perhaps due to the small sample size and different testing methodologies used by the PRS and BAPQ. In the future, it may be helpful to conduct this study with a larger sample and/or bring in a third pragmatic language measure.

Introduction

Pragmatic language, the language we use in different social interactions every day, is an essential life skill and the foundation for effective communication. However, mothers of children with an autism spectrum disorder (ASD) are at risk for impaired pragmatic language skills because of their increased likelihood (14-23% vs. 5-9%, the likelihood for mothers with typically developing children) to have the broad autism phenotype (BAP) [1, 2, 3, 4]. The BAP is a term used to describe the condition where a person displays symptoms similar to those found in an ASD but at a much milder level [3]. A trait of the BAP is impaired pragmatic language abilities [2, 5]. Mothers of children with Fragile X Syndrome (FXS) have also recently been found to have difficulties with pragmatic language because they either have FXS as well or the FMR1 premutation, an abnormality that occurs when there are 55-200 CGG repeats on the FMR1 gene [6]. Both the BAP and the FMR1 premutation are highly prevalent – 1 in 68 children in the U.S. have autism [7], and about 1 in 149 women have the FMR1 premutation [8]. In addition, a recent study has also shown that the pragmatic language traits of mothers with the FMR1 premutation is significantly correlated with the language development of their FXS children [9]. Thus, it has become increasingly important to have an accurate way of measuring pragmatic language in this adult population.

Two common measures of pragmatic language are the Pragmatic Rating Scale (PRS) [5] and the Pragmatic Language Subscale of the Broad Autism Phenotype Questionnaire (BAPQ) [10]. Both of these measures were developed to be used in adult BAP studies, with the former focusing more specifically on the pragmatic language component of the BAP and the latter aiming to serve as an efficient and reliable method for detecting the BAP in adults. The BAPQ's Pragmatic Language subscale questions were largely derived from the PRS [10], and it has since been found to have a detection rate of greater than 70% [3]. In the original study done with the PRS, parents of children with autism were shown to behave abnormally in terms of pragmatics more often than parents of typically developing children [5].

The current study, unlike previous ones that looked only at one measure, aims to compare these two measures of pragmatic language – the PRS and the Pragmatic Language Subscale of the BAPQ – to see how well they agree with each other in diagnosing pragmatic language abnormalities for mothers of children with ASD and mothers of children with FXS. It was hypothesized that the two measures would have high agreement at least in the mothers of children with ASD group, since both measures were designed to be used in adult BAP studies, and the Pragmatic Language Subscale of the BAPQ was designed with the PRS as the foundation. To determine the agreement, PRS and BAPQ data from a total of 76 mothers (mean age in years = 44.72, SD = 8.60) was reviewed. The mothers were recruited from ongoing studies on children with ASD or FXS at the University of South Carolina Neurodevelopmental Disorders Lab. Of the 76, 17 were mothers of children with ASD, 33 were mothers of children with FXS, and 26 were mothers of typically developing children (later referred to as the TYP group), and they were grouped as such. The TYP group was used as the control, and a cutoff score was determined for the PRS from its data. For the BAPQ, the cutoff score was taken from "The Broad Autism Phenotype Questionnaire: Prevalence and Diagnostic Classification" [3]. The mothers who scored above the cutoff for either measurement were categorized as "positive" for pragmatic language difficulties according to that measurement. Contrary to what was expected, across all three groups, there was little to no agreement between the mothers who tested positive for pragmatic language abnormalities on the PRS and the ones who tested positive on the Pragmatic Language subscale of the BAPQ.

Methods

This study specifically looked at the Pragmatic Rating Scale (PRS) and the Pragmatic Language subscale of the Broad Autism Phenotype Questionnaire (BAPQ), two common measures of pragmatic language for parents of children with an autism spectrum disorder (ASD). The goal was to determine how well these two measures agreed with each other in identifying mothers with pragmatic language abnormalities. To quantitatively determine which mothers had pragmatic language abnormalities, a cutoff score was used for both measures. The mothers with scores greater than the cutoff score were considered "positive" for abnormalities.

Pragmatic Rating Scale
The PRS was developed by Landa et al. in 1992 to help differentiate parents of children with ASD from parents of typically developing children. It includes 19 abnormal pragmatic behaviors, ranging from overly candid ("expresses very personal information or makes highly critical, evaluative comments about people or situations") to indirect ("overly subtle in expression of opinions or instructions with the result that the intended connotation or desired action is unclear"), that are to be rated on a scale of 0 to 2, with 0 being normal, 1 being moderately abnormal, and 2 being extremely abnormal [5]. The rating of each behavior is added up to determine the overall PRS score. For this experiment, two trained coders listened to a 30-minute conversation sample, watching for the 19 behaviors; the behaviors were then independently rated. Consensus scores were

subsequently determined by averaging the two ratings for each behavior, and the final rating (to be used in the data analysis) was rounded up if it ended up as 0.5 or 1.5.

A cutoff score was not provided in the literature for the PRS, so it was determined by repeating the procedure that had been used to determine the cutoff score for the Pragmatic Language Subscale of the BAPQ, which was given [3]. The mean score of all the TYP mothers was calculated, which turned out to be 5.731. Then, the standard deviation was calculated to be 3.424. This value was multiplied by 1.5 and then added to the mean to yield a cutoff score of 10.867. Any mother who scored higher than a 10.867 on the PRS was classified as positive for pragmatic language abnormalities.

Pragmatic Language Subscale of the Broad Autism Phenotype Questionnaire

The BAPQ is a self and informant report questionnaire, meaning that the subject fills out one copy of the questionnaire, assessing him or herself, and an informant, someone close to him or her (typically the subject's spouse) fills out another copy of the questionnaire assessing the subject. It was designed by Hurley et al. in 2007 to detect the Broad Autism Phenotype (BAP) in adults. Administered as the Personality Styles and Preferences Questionnaire (PSPQ) to control for dishonesty that might arise from knowledge of the questionnaire's purpose to diagnose the BAP, it has been shown to have a greater than 70% detection rate for the BAP [3]. The results of the BAPQ are divided into three subscales corresponding to the main components of the BAP: aloofness, rigidity, and pragmatic language. This study specifically looked at the Pragmatic Language Subscale score, which is calculated by averaging the responses of twelve of the questions on the questionnaire. The self-report copy of the questionnaire was filled out by each mother at the lab, while the informant-report questionnaires were mailed out with a self-addressed, stamped return envelope to reduce bias. If the informant filled it out on the spot, the answers given would have likely been influenced by the presence of the subject. Not all the informants mailed back the questionnaire however, so some mothers only had the self-report questionnaire on file. An average of the self and informant scores was taken and used as the best estimate scores for data analysis, but in the rare case where there were no informant scores, the self-report scores were used as the best estimate.

The cutoff score, 2.90, for the Pragmatic Language Subscale was taken from "The Broad Autism Phenotype Questionnaire: Prevalence and Diagnostic Classification" [3]. Any mother who scored higher than a 2.90 on the subscale was considered positive for pragmatic language abnormalities.

Results

The percentage of agreement between the mothers who tested positive for difficulties with pragmatic language on the PRS and on the Pragmatic Language subscale of the BAPQ was very low as seen in Figure 1. Positivity was determined using the cutoff scores shown in Table 1, where PRS and BAPQ scores above the cutoff were marked as positive. As seen in Table 2, only 5.88% of the mothers of children with ASD tested positive on both measures, followed by only 3.03% of the mothers of children with FXS, and 0% of the mothers of typically developing children. In total, only 2.63% of the full sample of mothers tested positive on both measurements.

Overall, the PRS determined that many more of the mothers were positive compared to the number determined to be positive by the Pragmatic Language subscale of the BAPQ in all of the groups except the TYP as shown in Figure 1. In the ASD group, the PRS found 23.53% (the sum of the percent positive on PRS only and the percent positive on both in Table 2) of the sample to be positive for pragmatic language issues, while the BAPQ only found 5.88% of them to be positive. Similarly, in the FXS group, the PRS found 42.42% of the sample to be positive, while the BAPQ only found 12.12% of the sample to be positive. The PRS and BAPQ found the same percentage of the TYP group to be positive (7.69%). For the full sample, 26.31% of the mothers tested positive on the PRS while only 9.21% tested positive on the BAPQ.

Discussion

Although it was predicted that the BAPQ and the PRS would have high agreement in identifying mothers who are positive for pragmatic language issues, that did not turn out to be the case. Rather, they agreed very little on which mothers were positive, and the PRS also identified a much larger percent of the mothers in each group (excluding the TYP) as positive compared to the percent identified as positive by the BAPQ. From the results, it is evident that the BAPQ and PRS are two very disparate measures. They share little to no agreement in determining which mothers are positive for the pragmatic language difficulties that are characteristic of the BAP and the FMR1 premutation [6]. The highest percent of agreements were in the ASD group, but even that was only 5.88%. This low agreement is especially surprising given how both of these tools were developed specifically to assess parents of children with ASD and how the Pragmatic Language Subscale of the BAPQ was developed using the PRS as a guideline. Beyond that, it is also rather unsettling that these two common methods of measuring pragmatic language, rather than supporting each other, appear more likely to contradict each other. However, this study was conducted with a fairly small sample size, and the ASD group had the lowest numbers (n = 17 while for the FXS group, n = 33 and for the TYP group, n = 26), so a future study done with a larger sample might yield different results.

The low agreement between these measures may also be related to the fact that the BAPQ and PRS are fundamentally different in their execution. The BAPQ's self and informant report nature might have been problematic due to rater bias. Participants are not always aware of their own tendencies, and while filling out the informant report, the participant's friend or spouse might have been hesitant to give poor ratings. Likewise, the PRS might have been problematic because a 30-minute conversation sample may not be long enough to accurately capture all of the participant's conversational habits. There is also a chance that being in a lab environment and recorded might have made the participant nervous or caused her to act abnormally. The differences between the execution of these two measures may also be the reason behind why the PRS classified so many more subjects as being positive compared to the BAPQ. Beyond this, the age difference of the two measures (the BAPQ is about 15 years younger than the PRS) may have also had an impact. To confirm these suspicions, more measurement studies, preferably with larger samples, would need to be conducted. It may also be useful to bring in other, more standard measures of pragmatic language that were designed for typical adults, such as the Pragmatic Protocol [11] and the Profile of Communicative Appropriateness [12], and see if those measures agree more with the BAPQ or the PRS.

Acknowledgements

Notes and References

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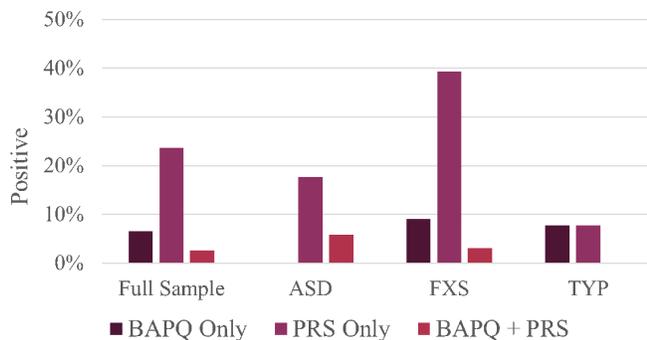
Figure 1. This shows the percentage of mothers who tested positive for pragmatic language issues for each group by the measure. The third bar shows the percentage of mothers who tested positive on both measures.

Table 1. The cutoff scores that were used to determine positivity for pragmatic language difficulties are shown. The BAPQ cutoff score used was the average of the self and informant report cutoffs for the Pragmatic Language subsection.

	PRS	BAPQ (PL)
Cutoff	10.867	2.900

Table 2. The number and percentage of mothers from each group who tested positive on the BAPQ, the PRS, or on both are shown. The TYP group served as the control group.

	Full Sample	ASD	FXS	TYP
No. Positive on BAPQ	5	0	3	2
No. Positive on PRS	18	3	13	2
No. Positive on Both	2	1	1	0
Total	76	17	33	26
% Positive on BAPQ	6.58%	0.00%	9.09%	7.69%
% Positive on PRS	23.68%	17.65%	39.39%	7.69%
% Positive on Both	2.63%	5.88%	3.03%	0.00%