PSYCHOLOGICAL FACTORS ASSOCIATED WITH GENETIC TEST DECISION-MAKING AMONG PARENTS OF CHILDREN WITH AUTISM SPECTRUM DISORDERS IN TAIWAN

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Making decisions to undergo Autism Spectrum Disorders (ASD) genetic testing can be challenging. It is important to understand how the perceptions of affected individuals might influence testing decision-making. Although evidence has shown that psychological factors are important in predicting testing decisions, affect-type variables have been largely ignored among well-established health theories. Using questionnaires, we examined genetic testing decision-making among 444 parents of children with ASD in Taiwan. The largest predictor of intention was anxiety (β=.46, p<.0001) followed by fear/guilt (β=-.041, p<.000). Attitudes did not predict intention. Affect-type variables should be considered in genetic testing decision making theory, research, and practice.

Introduction
As genetic technologies continue to advance in the post-genomics era, more genetic tests for Autism Spectrum Disorders (ASD) have been used to identify the causes of ASD, promote early detection and develop treatment plans. (Shen et al., 2010; Schaefer & Mendelsohn, 2013). The reasons might include the nature of this multifactorial condition (with a wide spectrum and different severity levels), the unclear clinical significance, the ambiguous interpretation of the test results, as well as a number of ethical, social, and legal questions pertaining to the test (Reiff et al., 2012; Marchant & Rober, 2009). Given these conditions, it is urgent to understand how affected individuals and their families view autism genetic testing and how their affective and cognitive perceptions might impact their decisions.

Albeit genetic tests are currently available for ASD-affected populations, making decisions to undergo autism genetic testing can be challenging for parents of children with ASD. It is important to understand how affected individuals and their families perceive autism genetic testing and how their perceptions might influence their decisions. Multiple lines of studies in various diseases have explored factors that might determine people's decisions associated with the uptake of genetic testing, such as perceived severity, perceived barriers and perceived benefits, attitudes and intention (Gooding et al., 2006). Although evidence has shown that psychological factors might be important in predicting genetic testing decisions, the affect-type variables have been largely neglected in the well-established health theories (Goodson, 2010; Gooding et al., 2006).

Although the reported prevalence of ASD in Taiwan (26.6 in every 10,000 person) was lower than the estimates from developed countries, a potential under-diagnosis and under-detection of ASD has made it an immediate public health concern (Sun et al., 2013; Lin et al., 2008). This phenomenon might be explained by the lack of awareness and recognition of ASD among clinicians, the lack of knowledge and acceptance of ASD in Taiwanese society, as well as the potential cultural influence in Taiwan (Sun et al.,

2013). For instance, parents might feel ashamed or embarrassed by having a child with ASD, which can lead to the difficulty in acceptance of the diagnosis. A few epidemiologic studies focusing on enhancing the detection rate of ASD among the Taiwanese population are being conducted. For example, one published study was designed to provide modified versions of ASD screening and diagnostic instruments (Wang et al., 2012). However, up till now, no recommended tests are available for patients and families with ASD in Taiwan. Since culture might have significant influence on Taiwanese families' perspectives toward genomic disorders and disabilities (such as eugenics and social stigma of having a child with birth defects), it is critical to examine the decision-making process with regard to ASD genetic testing before the provision of this test.

The purpose of this study is to address some gaps in the literature on the decisions to undergo autism genetic testing. This study explores the roles of 1) emotions, 2) attitudes, and 3) demographic variables in the decision-making process related to undergoing autism genetic testing among a sample of Taiwanese parents of children with ASD. Until now, no official recommendations are available for patients and families with ASD in Taiwan. Additionally, little is known about the factors determining the intentions to undergo autism genetic testing for parents of children with ASD in Taiwan.

Method
Sampling and recruitment
This study is a secondary data analysis of an international research project initiated by faculty at Texas A&M University and the National Hsinchu University of Education. The research team recruited parents of children with ASD through special education teachers enrolled in special education classes in Hsinchu area and Taoyuan County. The survey as well as information sheet were all distributed to potential parents by the teachers. This research obtained IRB approval from Texas A&M University. More details can be retrieved from our previous published work. (Xu & Lu, 2015)

Measures
Based on current literature about factors determining the attitudes, beliefs and decision making regarding genetic testing, (Gooding et al., 2006; Lerman & Crovle, 1996; Skinner et al., 2003) we established an integrative model for this study among a sample of Taiwanese parents of children with ASD (Figure 1).
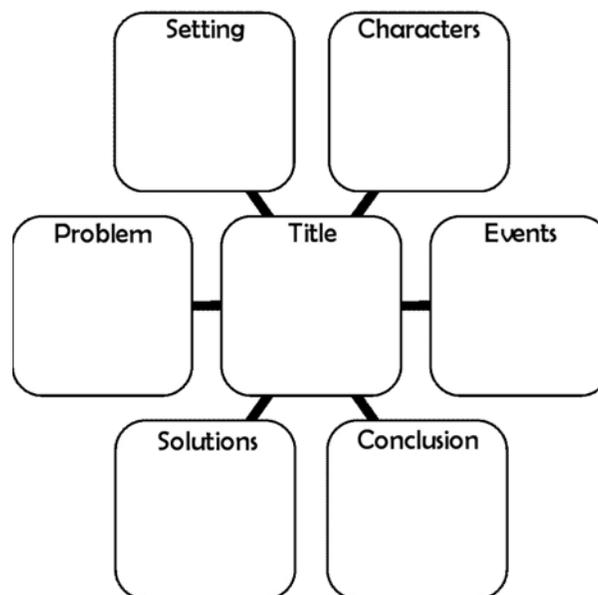
Figure 1. A Story Map (taken from Grünke, Wilbert, & Kim Calder Stegemann, 2013, p. 55)

Figure 1 The integrative model of emotions, attitudes and intention associated with undergoing autism genetic testing

Key constructs include four latent variables (anxiety, fear-&-guilt, attitudes, intention) while controlling for parents' age, gender, income, education, and religion. We also tested the five demographic factors (parents' age, gender, education, income and religious beliefs) as moderators. The variables in this model are from validated theories (Leventhal & Cameron, 2001; Ajen, 1991). The proposed model is designed to explain the emotional factors that facilitate or inhibit parents' decisions to undergo autism genetic testing. We added emotional factors as influences on the intention regarding autism genetic testing.

Data Analysis
We employed Structural Equation Modeling (SEM) analysis to assess whether the data supported the hypothesized model (Figure 1). Mplus 7.11 (Muthén & Muthén, 2007) was used to analyze the data due to its flexibility for handling different data structures and offering FIML (full information maximum likelihood) to handle missing data. The initial SEM step included a confirmatory factor analysis to establish a measurement model that determines latent model constructs. After establishing an adequate fit for the measurement model, we used the structural model to assess the underlying relationship between and among the proposed variables. To determine the fit between the hypothesized models and the observed data, we examined the following goodness-of-fit indexes: the chi-square, SRMR and RMSEA, based on the cutoff criteria-values of CFI more than 0.90, SRMR less than 0.05 and RMSEA less than 0.06.

Results
The final sample composed of 347 mothers and 97 fathers with autistic children in Taiwan. Parents' average age was 39.9 years (SD=5.4, range= 28-63). The average age of their spouses was 41.3 years (SD=5.6, range=26-63). The majority of the children involved (88%) in this research were boys diagnosed with ASD. The average age of these children is 9.5 (SD± 2.24). About 67.3% of the parents' education level was below college and 74.4% had an annual household income less than $40k.

Structural model
After confirming through factor analysis that the measurement model exhibited appropriate fit, we performed SEM to verify the proposed structural relationships in this study. First, we sought to understand if the proposed model is adequate for explaining parents' intentions to undergo autism genetic testing. This model suggested that the proposed model offered an adequate explanation of the observed empirical data. Based on our results, we found that the more anxiety parents have, the more likely they are to pursue the test. However, with more fear or guilt, the parents were more likely to decline the test. Although a large percent of the parents had positive attitudes toward the test, attitudes did not appear to predict parents test intention.

Second, we sought to assess if the proposed model exhibits different patterns and values depending on participants' demographic characteristics. We also examined the structural variation of anxiety, fear & guilt, attitudes and intention using age, gender, education, income, and religion as moderators. In order to verify the structural invariant, we used MLR estimators to simultaneously assess both the unconstrained and unconstrained models. The structural paths were equally restricted across dichotomized groups, i.e. age≥ 35 years and < 35 years, male and female, high and low income (≥ 40K, <40K), high education and low education (college graduates or below college), as well as with religious beliefs and without religious beliefs. However, none of the moderating effects of age, gender, education and income yielded Satorra-Bentler scaled chi-square value. The diagnosis from the output indicated no convergence due to exceeded interactions. These phenomena might be caused by the sparse of data for the dichotomized groups. Therefore, we were not able to infer whether age, gender, education, income, and religion interacted significantly with the latent variables.

Lastly, we were interested to know which variables in the proposed model are the best predictors of parents' intentions to undergo genetic testing for ASD. In the proposed model, the largest predictor of

intention was anxiety (β=.46, p<.0001). In addition, fear and guilt were also predictors (β=-.041, p<.0001). The use of the squared multiple correlation (R^2), the percentage of variance explained by one or more predicator variables on a dependent variable-in SEM is an ongoing area of research and a trend in explaining social phenomena. According to the model results, about one tenth of variance in parents' test intention can be explained by anxiety, fear and guilt.

Discussion

The purpose of this study was to utilize structural equation modeling analysis to examine the associations between the emotions, attitudes and intentions, as well as test the overall "fit" of the proposed model in this study. Our findings extend existing literature on decision making about undergoing genetic testing for ASD in two ways.

First, we used an integrative model and SEM analysis to understand how emotions and attitudes might influence parents' intentions to undergo ASD genetic testing. Previous literature have used validated health theories for understanding the factors predicting intentions toward genetic testing, (Johnson et al., 2011); however, emotional factors have not been adequately addressed in these theoretical frameworks (Buhi et al., 2011). Notably, this study answers the call from the National Health Genomics Research Institute to expand beyond the existing conceptual models for exploring stronger predictors of genetic test decisions (Wade et al., 2012). We added affect-type variables, a largely overlooked factor in genetic testing decisions, as key constructs in our proposed model.

It is noteworthy that our findings demonstrated the negative influence of fear and guilt on parents' intentions to undergo ASD genetic testing. Past studies have shown that fear or guilt might potentially lead to a decline in genetic tests or refusal to participate in genetic research (Shumacker et al., 1996; Chen & Goodson, 2007). The particular kind of fear or social stigma in this study was associated with social, legal and ethical concerns related to testing. In addition, we specifically measured guilt caused by passing the ASD-associated genes onto the affected child and guilt caused by undergoing genetic testing. The specific kinds of fear and guilt we assessed, might be attributed to the culture and societal factors related to having a child with genetic disorders (Yuan & Bentler, 2010; McBride et al., 2008, Aatre & Day 2011). Similar to other Asian societies, the Taiwanese society might also demonstrate discriminatory attitudes toward people with disabilities, particularly, with mental illnesses (Eisenbruch et al., 2004; Yang et al., 2013).

A second way that our findings contribute to the existing literature for genetic testing is by having direct implications for public health genomics education and practice. The proposed model suggests that, educational interventions might be important based on the identified relationships among the factors. Although our sample did not allow us to generalize to the entire Taiwan population, our study provided support to the need of pre-test counseling as well as genomic or genetic education tailored to parents of children with ASD and parents with other genetic disorders among the general public in Taiwan. There is a significant literature gap in the studies that have explored the associations between emotional factors and the genetic test decisions, as most studies have only focused mainly on the attitudes and intentions related to genetic testing. Our study helped to close the gap by providing theory-based evidence specifically addressing the emotions, attitudes and intentions among a sample of 444 parents of children with ASD in Taiwan. Furthermore, our study also confirmed anxiety, fear and guilt were associated with parents' test intentions; attitudes did not appear to be an influencing factor in the decision-making process. One of the most profound findings in this study is the proposed framework which better reflects empirical evidence and encourages re-integration of theories to explain decision-making processes related to genetic testing.

Conclusion

Limitations

Several limitations of this study deserve attention when interpreting the results from this study. First, this secondary analysis was from a cross-sectional study among children in public schools in Taiwan. Thus, the sample might not adequately represent responses from autistic children's parents from other settings, for instance, private schools or home schools. Another limitation was that this study assessed the influence of anxiety, fear and guilt and attitudes on intention. However, perceived recurrence risks and perceived severity levels might also have influence on parents' intentions to undergo genetic testing for ASD. Therefore, these factors might need to be examined further.

Implications

This study is useful in providing insights about parents' perspectives regarding ASD genetic testing prior to the full implementation of this test in Taiwan. It will also contribute to the development of better genetic services and research in Taiwan. Our findings also suggest that it is important to design more culturally appropriate educational programs for parents of children with ASD in Taiwan. Most participants postulated favorable attitudes toward ASD genetic testing and were likely to take their affected children for ASD genetic testing. However, compared to the situation in the US, where ASD genetic testing is offered as a routine health care service, there is a lack of valid and reliable ASD genetic testing procedures in Taiwan. Given that the parents of children with ASD were interested in ASD genetic testing, there is an immediate need to develop valid and reliable testing methods in Taiwan and provide culturally appropriate pre-test educational interventions for parents of children with ASD.

Future studies on decisions about undergoing genetic testing can benefit from addressing gaps in research pointed out by this study. More studies are needed to 1) examine emotions related to the intention to undergo genetic testing for ASD among the affected populations, 2) test theories that integrate emotions and other under-investigated factors such as perceived barriers, perceived benefits, and social environmental factors associated with the test intention; 3) evaluate moderation effects of demographic information. Future studies will also benefit from addressing methodological quality dimensions, such as the employment of more rigorous designs and the use of comparison groups. Addressing these dimensions is crucial for achieving a clearer understanding of parents' intention with regard to genetic testing for ASD for ASD-affected families and communities in a wider range of geographical locations.

Despite the limited generalizability, our findings also inform communications related to ASD genetic testing among health-care providers and parents of autistic children. For instance, in order to provide better genetic services for families affected with ASD, the multi-disciplinary medical team involved in the diagnostic process of ASD, such as pediatricians, medical geneticists and neurologists should proactively consider the possible distress among parents of children with ASD. Since healthcare providers or health education specialists can play a critical role in shaping people's decision to undergo genetic testing, it is also important to educate them and enhance their knowledge so that they can help parents make more informed decisions regarding undergoing autism genetic testing. Furthermore, to manage parents' concerns resulting from fear, policymakers and legislators also need to consider genetic discrimination laws in Taiwan.

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