

Child and Parental Factors Affecting Coping Strategies and Psychosocial Outcomes for Parents
with Children with Prader-Willi Syndrome

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Abstract

Prader-Willi Syndrome (PWS) is a rare, genetic neurodevelopmental disability characterized by hyperphagia, mood swings and intellectual disability. Families with a child with PWS often experience increased family tensions and a greater number of stressors, even compared to families with children with different intellectual and developmental disabilities (IDDs). However, there is variation in parental stress, anxiety and depression levels. This study explored the effects of both child factors, such as hyperphagia and severity of behavioral problems, and parental factors, such as coping strategies, on parental stress, anxiety and depression, for parents of children with PWS (n = 154). Results show that child factors significantly impact parental stress, anxiety and depression. Parental usage of rational, active coping strategies is strongly linked to lower stress and depression levels, while parents who employ avoidant, helplessness coping strategies are more likely to have higher levels of stress, depression and anxiety. While there were no significant interaction effects between parental coping strategies and the severity of the child's behavioral issues on parental stress levels, the best outcomes were achieved when parents did not use avoidant coping strategies and their children had fewer behavioral problems. Future research implications and outcomes are discussed.

Introduction

Prader-Willi Syndrome (PWS) is a rare genetic neurodevelopmental disorder occurring in about 1 in 25,000 births as a result of specific genetic abnormalities on chromosome 15 (Laurier et al., 2014). PWS is characterized by hypotonia, hyperphagia, intellectual disability, skin picking, mood swings and other behavioral challenges (Laurier et al., 2014). Researchers have examined many factors affecting the behavioral problems that present with PWS, such as

discrepancies between genetic subtypes. There are multiple genetic causes of PWS, all affecting the paternally expressed genes on chromosome 15q11–q13, with most of the cases resulting from parental deletion (65–70 %) or maternal uniparental disomy (mUPD; 20–30 %) (Cassidy, Schwartz, Miller, & Driscoll, 2012). There are significant behavioral differences between the deletion subtype and the maternal uniparental disomy subtype, with the mUPD group being at a higher risk for Autism spectrum disorders (Veltman, Craig & Bolton, 2005). These specific between-group variations are still being researched, but the differences in genetic subtype have been demonstrated to have a significant impact on behavior (Veltman, Craig & Bolton, 2005). For example, the mUPD group has been demonstrated to have higher verbal abilities, decreased skin picking, increased rectal-picking and decreased facial recognition abilities than the deletion subtype (Roof, Stone MacLean & Feurer, 2000; Dykens & Roof 2008; Key, Jones & Dykens, 2013). There are many chronic behavioral issues that carry over from childhood into adulthood, with age being negatively correlated with problematic behaviors and moodiness for people with the Type I deletion, which places parents in a state of constant stress throughout their child's lifetime (Dykens & Roof, 2008).

Parents of children with PWS often experience higher levels of daily stress than parents of typically developing children as a result of their familial situations, as well as compared to families of children with other intellectual and developmental disabilities (IDDs); this negatively affects the behaviors of both the child and the parents (Tvrđik et al., 2015). The effects of this chronic stress are devastating, negatively impacting parents' physical and mental health (Arnsten, 2009). Chronic stress causes catastrophic effects on the brain, weakening the prefrontal cortex, which regulates stress levels through negative feedback, and strengthening the amygdala, which stimulates the body's stress response; these changes decrease overall cognitive

functioning (Arnsten, 2009). Furthermore, it was found that compared to parents with typically developing children, parents of children with IDD experienced overall worse health outcomes, which were strongly positively correlated with high stress levels (Cantwell, Muldoon & Gallagher, 2014).

When compared to families of children with Williams Syndrome and Fragile-X Syndrome, families with children with PWS experience more family stress, also correlated with higher levels of marital conflict and parental anger; this is associated with an increased amount of reported behavioral issues in the children (van Lieshout, Meyer, Curfs, & Fryns, 1998). When there was more parental anger, the children with PWS were less agreeable, less conscientious, less emotionally stable, less open and more irritable (van Lieshout, Meyer, Curfs, & Fryns, 1998). This relationship is bidirectional, creating a dangerous cycle as parents' increased anger leads to more negative behaviors in their children, thereby reinforcing the stress of the parents (van Lieshout, Meyer, Curfs, & Fryns, 1998).

The relationship between parental stress levels for PWS families is mediated by a number of factors. The utilization of effective coping methods has been demonstrated to decrease parental stress levels (Tvrdik et al., 2015). Parents are also shown to be negatively impacted by the number of negative behaviors demonstrated by their child, reporting higher levels of stress when their child demonstrates a greater number of behavioral problems, and with stress levels remaining unaffected by the child's level of cognitive abilities, age or degree of obesity (Hodapp, Dykens & Masino, 1997). As shown by Lanfranchi & Vianello (2012), parental stress levels are affected by factors outside of their control, which makes it more difficult for them to cope with the stressors. In a comparison between stress levels and locus of control among parents of children with Down Syndrome, Williams Syndrome, Fragile X Syndrome and PWS, the PWS

group had the highest stress levels and also demonstrated an externalized locus of control, in which they felt that an outside force had control over their lives, as a result of the increased severity of behavioral problems with PWS (Lanfranchi & Vianello 2012). Furthermore, although both parents of children with PWS demonstrated a more external locus of control than the comparison groups, the mothers reported stronger feelings that their PWS child dominated their lives compared to the fathers, theorized to be the result of many mothers spending more time with the children in their daily lives (Lanfranchi & Vianello 2012).

Compared to families with children with complex health conditions, parents and siblings of children with PWS were more likely to experience worse family functioning, communication problems, depression and increased feelings of isolation, anger and worry (Mazaheri et al., 2013). It is difficult for parents to have time to themselves because individuals with PWS have so many distinct behavioral challenges, such as hyperphagic food-seeking behaviors; the children require almost constant attention (Tvrđik et al., 2015). Parents must be hyperaware of their child's actions for health and safety reasons, as some people with PWS can literally eat themselves to death if unattended (Stevenson et al., 2007). More research is necessary to develop programs to benefit this neglected parental population and to improve their quality of life by reducing their long-term stress levels (Collins & Schwartz, 2011; Tvrđik et al., 2015).

As a result of consistent behavioral challenges facing both the child with PWS and their parents, these caregivers face a unique set of challenges to their own well-being (van Lieshout, Meyer, Curfs, & Fryns, 1998). The health of primary caregivers of individuals with IDD is often forgotten amidst the multi-faceted, all-encompassing needs of the person with special needs that they are caring for (Collins & Schwartz, 2011). Doctors, therapists and other professionals usually focus most of their attention and concern on the person with the disability,

because those individuals require unique care and have a wide array of obvious social, behavioral and physical needs (Collins & Schwartz, 2011). As a result of spending so much time looking after their child, caregivers' own health can fall by the wayside as they neglect their self-care, and parents can find themselves facing a decreased quality of life as a result (van Lieshout, Meyer, Curfs, & Fryns, 1998). There is a wide range of self-reported life satisfaction for mothers of children with PWS, with both child factors, such as behavioral issues, and maternal factors, such as stress, exerting significant effects on maternal quality of life (Shivers, Leonczyk, & Dykens, 2016).

Not exclusively in PWS, parental roles, status and personality were all correlated with different coping mechanisms for families with children with IDD; for example, parents scoring higher in neuroticism were more likely to develop maladaptive coping methods (Glidden, Billings & Jobe, 2006). The COPE questionnaire has often been used to examine parental coping mechanisms, dividing coping behaviors into three categories: Emotion-Focused Coping, Rational/Active Coping, and Avoidance/Helplessness Coping (Lyne & Roger, 2000). However, in mothers with children with PWS, emotional coping has not been shown to significantly affect quality of life, whereas usage of rational coping strategies is positively correlated with levels of life satisfaction and usage of avoidant coping strategies is negatively correlated with levels of life satisfaction (Shivers, Leonczyk, & Dykens, 2016). For typically developing children, mothers experiencing higher levels of stress during their child's adolescence are more likely to frequently utilize avoidant coping strategies, in a unidirectional relationship (Steger, Gondoli & Morrissey, 2013). These avoidant coping strategies are related to increases in depressive symptoms, but prior experiences with depression can also prime a mother to engage in less constructive and

more avoidant coping strategies; there is a bidirectional relationship between depressive symptoms and avoidant coping (Steeger, Gondoli & Morrissey, 2013).

In an intervention study focused on the development of beneficial coping mechanisms for parents of children with PWS, parents demonstrated more coping behaviors in general after the intervention, with a specific increase in the utilization of coping techniques that were related to “maintaining family integration” than before the trial, coinciding with a significant decrease in parental stress levels (Tvrdik et al., 2015). This intervention trial also demonstrates the significance of this research, as discovering possible mediating variables for parental stress levels could lead to the development of more effective educational programs about beneficial coping strategies for parents of children with PWS, that could possibly be applicable for parents of children with other IDD's (Tvrdik et al., 2015).

Despite all of these findings, providers do not place enough emphasis on the needs of the caregivers, typically the parents, who are also experiencing unique difficulties and in need of additional support (Collins & Schwartz, 2011). There are so few intervention programs focused on helping parents of children with IDD's, much less PWS specifically, despite the evident need (Tvrdik et al., 2015). Furthermore, it may be difficult for parents to seek help when they have so little time to themselves and they are constantly worried about putting the well-being of their family before their own needs (Collins & Schwartz, 2011). In addition to the negative impacts on cognition from experiencing higher levels of stress in general, parents of children with PWS are also more at risk for a variety of mental health problems, with higher rates of somatization, phobic anxiety, obsessive-compulsive and anxiety problems in comparison to the control group (Skokauskas, Sweeny, Meehan & Gallagher, 2012). In general, mothers of children with IDD's are at greater risk of depression, with family strain, parental stress and family income being the

key factors responsible for this augmented risk (Azar & Badr, 2006). While there is a plethora of information regarding stress levels for parents of children with PWS, little to no research specifically focused on parental mental health problems has been conducted (Skokauskas, Sweeny, Meehan & Gallagher, 2012). More research on these mental health concerns is necessary, because there has been a shift away from unidirectional models of stress associations with mood disorders; other factors such as environmental context and personal characteristics impact the development of mental health concerns (Hammen, 2005).

In this paper we will examine the following five research questions:

1. How are unique components of PWS, such as hyperphagia and genetic subtype, related to parental stress and the severity of child behavioral issues?
2. How are parental psychosocial outcomes affected by their children's behavioral issues?
3. For parents with children with PWS, what are the connections between stress and mood disorders, such as depression and anxiety?
4. What is the relationship between parental coping strategies and mood disorders, such as depression and anxiety?
5. How are parental coping strategies related to child factors, such as behavioral issues and hyperphagia?
6. How do the effects of both parental coping strategies and child behavioral issues impact parental stress levels?

Methods

Participants

The participants were enrolled in an ongoing longitudinal study, Trajectories of Phenotypic Behavior in Prader-Willi Syndrome, at the Vanderbilt Kennedy Center. All participants had a confirmed genetic diagnosis of PWS. The data set includes 154 participants' reports during their first visit completing the 2001 Child Behavior Checklist (CBCL) over the course of 2009 to 2014.

Parental measures indicated that the data set was skewed to represent a majority of married parents, with one other child in addition to their child with PWS, and an income of more than \$100,000 (married marital status = 75.3%; one other child = 40.9%; household income > \$100,000 = 42.9%). These three key demographic factors (marital status, number of other children and household income) are often considered to be three potentially moderating factors for parental stress in families with a child with an IDD.

The ages of the children ranged from 4 to 55; the average age was 14.9. The children's gender breakdown was split fairly evenly; 44.2% of the sample was male and 55.8% of the sample was female. There were more participants presenting with the deletion genetic subtype than the mUPD genetic subtype, (deletion = 64.9% and mUPD = 35.1%) consistent with the occurrence rate in the general PWS population (Cassidy, Schwartz, Miller, & Driscoll, 2012). The participants were primarily Caucasian (86.3%). Full demographic information can be found in Table 1.

Procedure

All participants in the data set completed a minimum of one visit, with some coming in for as many as four visits total. During the course of each site visit the caregivers completed assessments about their child's behaviors and health, as well as their own well-being and family stress, with one researcher. Simultaneously, another research completed the other assessments

with the child. However, for continuity, the only data used for the purpose of this study came from the first visit in which the 2001 CBCL was completed. All of the data obtained for this study was collected from 2009-2014. Researchers obtained informed consent and assent from all participants included in the study.

Measures

Demographics

Demographic variables included child age, child genetic subtype (mUPD or deletion), child gender, child race, number of other children in the family, parental marital status and household income.

Measures of the Child

Behavioral Issues: The Child Behavior Checklist (CBCL; Achenbach and Rescorla 2001) was used to assess behavioral problems in the child with PWS. The parents answered questions about their children's behaviors on a 3-point scale (0 = not true, 1 = sometimes or somewhat true, 2 = often true). The summation of these scores are standardized by age and gender to provide the final T-score encompassing total behavior problems. Higher scores signify more severe behavioral issues. A t-score of 0 to 64 is considered to be in the normal range, a t-score of 65 to 69 is considered to be in the borderline clinical range, and a t-score of 70 or above is considered to be in the critical range. The CBCL has been used in the past to assess individuals with PWS (Dykens, & Lambert, 2013; Shivers, Leonczyk, & Dykens, 2016).

Hyperphagic Behaviors: The extent of hyperphagic behaviors in the child with PWS was assessed using the 13-item Hyperphagia Questionnaire (Dykens et al., 2007). This study used a modified version of the measure, in which parents answered a 10-item inventory, rating their child's hyperphagic behaviors (e.g. "How often does your child try to steal food") and

hyperphagic drive (e.g. “How upset does your child generally become when denied a desired food”) on a scale of 1-5 (1 = not a problem to 5 = a severe or frequent problem). They also reported the age when their child began to show an increased interest in food as well as the variability in their child’s drive for food. The summation of these scores resulted in the raw total hyperphagia score which was utilized for analysis.

Measures of the Parent

Parental Anxiety: The Beck Anxiety Inventory (BAI; Beck, Epstein, Brown, & Steer, 1988) is a 21-item self-report measure of anxiety in which participants are asked to what extent they were affected by specific symptoms of anxiety over the past month (e.g. “Fear of losing control” and “Wobbliness in legs”). All items are answered on a 4-point Likert scale, from 0 to 3 (0 = not at all to 3 = severely; could barely stand it). The summation of the scores for each question result in the final raw score, which was used for analysis. The scores can range from 0 to 63. A score of 0–9 indicates normal or no anxiety; 10–18 indicates mild to moderate anxiety; 19–29 indicates moderate to severe anxiety; and 30–63 indicates severe anxiety. It was originally created to discriminate between anxiety and depression.

Parental Depression: The Beck Depression Inventory (BDI; Beck et al., 1961), a 21-item self-report measure, was used to gauge the presence and severity of depression. Each question focuses on a specific symptom of depression (e.g. Pessimism, worthlessness, sadness) and is answered on a 4-point Likert scale, from 0 to 3 (e.g. 0 = “I do not feel sad” to 3 = “I am so sad or unhappy that I can't stand it”). The summation of the scores for all of the items provides the final raw score to be used for analysis. The scores can range from 0 to 63. A final score of 0-13 is in the minimal depression range; 14-19 is in the mild range; 20-28 is in the moderate range; and 29-63 is in the severe range.

Parental Stress: A short version of the Parenting Stress Index (PSI; Abidin, 1983) was used to measure parental stress. The assessment involved 36 self-report questions answered on a 5-point Likert scale (1 = strongly agree to 5 = strongly disagree). All of the questions fell into one of three categories: Personal Distress (e.g. “I feel trapped”), Parent-Child Dysfunction (e.g. “My child rarely makes me feel good”) or Difficult child (e.g. “My child is very moody and easily upset”). The summation of these scores provides the final Total Stress score, which was used for analysis. The measure is regarded as having reliability, internal consistency and validity (Abidin, 1995). It has been used to assess mothers of children with PWS in past studies (Shivers, Leonczyk, & Dykens, 2016).

Parental Coping: A short form of the COPE questionnaire (Carver, Scheier, & Weintraub, 1989) was used to evaluate parental coping strategies. The measure is made up of 52 self-report items that are all evaluated on a four-point Likert scale (1 = usually don't do this at all to 4 = usually do this a lot). This study analyzed three coping factors: Rational, active coping (e.g. “I try to come up with a strategy about what to do”), Emotional coping (e.g. “I feel a lot of emotional distress and I find myself expressing those feelings a lot”) and Avoidant, Helplessness coping (e.g. “I act as though it hasn't even happened”). The questionnaire provides three separate scores, one for each of the three coping factors; each score is the result of a summation of the questions for a specific coping factor. The COPE has been used to assess mothers of children with (IDDs) in the past, as well as specifically mothers of children with PWS (Seltzer, Greenberg & Krauss, 1995; Woodman & Hauser-Cram, 2013; Shivers, Leonczyk, & Dykens, 2016).

Results

Hyperphagia, CBCL and PSI

Bivariate correlation analyses demonstrated significant relationships between various components of PWS that could impact parental stress levels, as well as the cyclic nature of other negative behaviors. There was a strong significant correlation between children's hyperphagic behaviors and parental stress levels ($r = .404, p = .000$), as well as between children's general behavioral issues and parental stress levels ($r = .637, p = .000$). Furthermore, there was a strong relationship between children's hyperphagic behaviors and their overall behavioral issues ($r = .456, p = .000$).

Genetic Subtype with Stress and CBCL

One-way ANOVA analyses indicated that while there was a definite trend, there was not a statistically significant difference between deletion and mUPD genetic subtype groups for parental stress ($F = 2.937, p = 0.089$) or children's behavioral issues ($F = 2.876, p = 0.092$). While children with the mUPD subtype ($n = 54$) did have higher mean CBCL scores (65.130) and parents had higher mean stress levels (91.611) than for children with the deletion subtype ($n = 100, CBCL: 62.560, PSI: 85.570$), significant between-group differences for the mUPD and deletion genetic subtypes were not observed.

Children's Behavioral Issues with Parental Outcomes

Using linear regression analysis to examine the effects of children's behavioral problems on parental psychosocial outcomes, there were significant relationships between children's CBCL scores and parental stress ($r^2 = .406, B = 1.483, p = .000$), anxiety ($r^2 = .152, B = .242, p = .000$) and depression levels ($r^2 = .172, B = .392, p = .000$). There was a very strong relationship between stress levels and the children's negative behaviors, but the parental depression and anxiety levels could not be explained by their children's behavioral issues alone.

Parental Stress and Mood Disorders

Using linear regression analysis to examine the effects of parental stress on parental psychosocial outcomes, there were significant relationships between stress levels and parental anxiety ($r^2 = .210$, $B = .122$, $p = .000$) and parental depression ($r^2 = .353$, $B = .242$, $p = .000$). While these relationships were significant, and parental stress does have an impact on parental levels of anxiety and depression, stress did not account for the entirety of these mood disorders, as would be expected by multidirectional models of depression and anxiety.

Parental Coping and Mood Disorders

Using linear regression analysis to examine the effects of parental parental coping on parental psychosocial outcomes, parental coping strategies had significant effects on stress and depression. There was no significant relationship between rational coping and parental anxiety ($R = -.063$., $r^2 = .004$, $B = -.045$ $p = .436$), but higher rational coping scores were associated with lower depression levels ($R = -.204$., $r^2 = .086$, $B = -.318$ $p = .000$). Higher avoidant coping strategy scores were significantly related to both higher levels of parental anxiety ($R = .218$., $r^2 = .048$, $B = .326$ $p = .007$) and parental depression ($R = -.063$., $r^2 = .004$, $B = -.045$ $p = .436$).

For both the rational and avoidant coping factors, participants were divided into two group based on their scores, with the bottom 75% of scores being ranked into the “Low” group and the upper 25% of scores being ranked into the “High” group. The ranges for the data set, for both rational and avoidant scores, did not encompass the full range of scores available for the questionnaire (Rational: Total Possible Range = 18-72 and Experimental Range: 36-72; Avoidant: Total Possible Range = 11-44 and Experimental Range: 12-31). In addition, there was no standardization of the measure to assess what qualified as a “low” or “high” score, so to create these groups, the researchers concluded that participants with scores up to one standard deviation above the data set’s mean would fall into the “Low” group; all participants with scores

equal to or greater than one standard deviation above the mean were in the “High” group

(Rational: Mean = 55.444, Low Group = 18-61, High Group = 62-72; Avoidant: Mean = 17.575, Low Group = 11-20, High Group = 21-31). Using one-way ANOVA analysis, researchers examined potential between-group differences on measures of parental stress, anxiety and depression for these “Low” and “High” rational and avoidant groups.

For the rational coping factor groups (Low: N = 114, High: N = 40), results indicated that there were moderate, significant between-group differences for parental stress and depression, with the “High Rational” group exhibiting lower levels of stress and depression (PSI: $F = 5.050$, $p = .026$; BDI: $F = 7.309$, $p = .008$). However, while higher rational coping scores were associated with lower anxiety scores, there were not significant between-group differences when looking at parental anxiety levels (BAI: $F = 1.842$, $p = .177$).

Examining the avoidant coping factor groups (Low: N = 125, High: N = 29), there were large, significant between-group differences for parental stress and depression; the “High Avoidant” coping group exhibited higher levels of stress and depression (PSI: $F = 19.807$, $p = .000$; BDI: $F = 22.212$, $p = .000$). Although higher avoidant coping scores were associated with higher anxiety, there were not significant between-group difference for anxiety levels (BAI: $F = 2.889$, $p = .091$).

Parental Coping and CBCL/Hyperphagia

Bivariate correlation analyses of the relationships between behavioral issues, hyperphagia and the different coping factors produced a variety of results. As previously discussed, there was a strong, significant relationship between hyperphagic issues and overall behavioral problems ($r = .456$, $p = .000$). When examining parental rational coping strategies in conjunction with child factors, there was a small, non-significant negative correlation between behavioral

issues and rational coping ($r = -.134, p = .098$). However, in contrast, there was a small, non-significant positive correlation between hyperphagia and rational coping ($r = .053, p = .519$). For parental avoidant coping strategies, there were moderate, significant positive correlations for both behavioral issues ($r = .355, p = .000$) as well as hyperphagia ($r = .352, p = .000$).

Between-Group Analyses of Parental Coping and CBCL

Using the same “High” and “Low” groups for parenting styles, Univariate analyses of variance were conducted also using “High” and “Low” groups for children’s behavioral issues in order to examine both parent and child factors affecting parental stress levels. While the parenting coping factor groups were based on the data set’s specific means, the behavioral issues groups were determined according to CBCL standardizations. A CBCL score equal to or greater than 65, encompassing both the borderline and critical scores, would place the child into “High CBCL” group; a score of 64 or below would place the child into the “Low CBCL” group (High: $N = 77$, Low: $N = 77$).

When examining parental stress levels in relation to child behavioral issues and parental rational coping strategies, there were strong, significant differences between the high and low groups for both variables (CBCL: $F = 28.551, p = .000$; Rational Coping: $F = 4.249, p = .041$). However, there were no significant interaction effects between CBCL groups and rational coping groups (CBCL Groups * Rational Groups: $F = 1.540, p = .216$). Graph 1 demonstrates the between-subject effects of the groups: the “High Rational” parental group always exhibited lower stress levels, despite CBCL scores, than the “Low Rational” group. Across both the “High Rational” and “Low Rational” groups, the “Low CBCL” group demonstrated significantly lower stress levels than the “High CBCL” group. Therefore, for these comparisons, the lowest parental

stress levels were reported for parents who utilized more rational coping strategies and whose children had the lowest severity of behavioral issues.

Parental stress levels were also examined while looking at child behavioral problems and parental avoidant coping strategies. These results also demonstrated very strong, significant differences in parental stress levels for the different CBCL and avoidant coping groups (CBCL: $F = 19.325$, $p = .000$; Avoidant Coping: $F = 13.984$, $p = .000$). There were no significant interaction effects between the two variables (CBCL Groups * Avoidant Groups: $F = .334$, $p = .564$). Graph 2 shows the effects of these variables on parental stress levels. The “High avoidant” parent group always exhibited higher stress levels than the “Low avoidant” stress group, regardless of the severity of their children’s behavioral problems. For both the “High Avoidant” and “Low Avoidant” groups, the parents whose children were in the “Low CBCL” group had lower stress levels than those whose children were in the “High CBCL” group. As a result, the parents who utilized fewer avoidant coping strategies and whose children had the least severe behavioral issues reported the least parental stress.

Comparing the Univariate analysis results for rational and avoidant coping strategies, there were much stronger, more significant effects on parental stress for avoidant coping strategies than there were for rational coping strategies (Rational Coping: $F = 4.249$, $p = .041$; Avoidant Coping: $F = 13.984$, $p = .000$). However, for parents with higher rational coping scores, the effects of their children’s behavioral issues had a more significant effect on their stress levels (CBCL for Rational Coping: $F = 28.551$, $p = .000$; CBCL for Avoidant Coping: $F = 19.325$, $p = .000$). Neither of the coping strategies had interaction effects with the children’s behavioral issues, demonstrating the effect of at least one moderating variable, as explored in the general discussion.

General Discussion

This study examined the various factors, both child and parental, affecting parental psychosocial outcomes for parents with children with PWS. This is under-researched, underserved population, with the majority of research about PWS being conducted on the individuals with PWS; there is not enough focus also placed on the well-being of their caregivers. Because parents who have a child with PWS have been shown to have higher stress levels than parents of children with other IDD's, the researchers sought to determine what specific factors had the biggest impact on parent stress (Tvrdik et al., 2015). As expected, there was a moderate relationship observed between global behavioral issues and hyperphagic behaviors, with the two factors being intertwined. The results also indicated that both hyperphagic behaviors and the severity of overall behavioral issues has strong, significant relationships with parental stress level. There was a stronger association between parental stress and global behavioral problems, which was consistent with common, informal parental reports. Parents often express that their children's compulsive behaviors and mood swings are the most difficult, stressful part of the disability, with food issues being secondary, and these findings support their claims.

Genetic subtype is another important child characteristic thought to be related to differences in behavioral problems, as well as an important factor affecting parent's stress. Surprisingly, this study did not find significant differences between the deletion and mUPD groups for parental stress levels or overall behavioral issues; the researchers predicted that there would be bigger between-group differences. While there was a general trend, with participants expressing the mUPD subtype having more severe behavioral problems and their parents reporting higher levels of stress, these relationships were not significant. The researchers theorize

that although there are specific phenotypic differences between the groups, the measures used to assess the participants might not have been able to capture the specific incongruities between the two genetic subtypes. The CBCL and PSI are both global measures and not specific to PWS. Although both have commonly been used to assess individuals with PWS in other studies, neither of them have items related to several key components of what makes PWS such a unique neurodevelopmental disorder. For example, although the CBCL captures some of the compulsive behaviors associated with PWS (e.g. rating the prevalence of impulsive behaviors and obsessive thoughts), neither inventory mentions food-seeking behaviors, which are key components of the behavioral challenges that accompany a PWS diagnosis.

In addition to the observed effects of children's behavioral issues on parental stress levels, the severity of their child's behavioral problems also had strong, significant effects on parental anxiety and depression levels. As well, examining internal, parental factors, there was a strong, highly significant relationship between the effects of stress on depression and anxiety levels, with stress having a slightly stronger effect on anxiety levels. To the researchers' knowledge, this is the first study that has examined specific child factors contributing to anxiety and depression in parents with a child with PWS. While past studies have discovered that these parents are at higher risk for anxiety and depression, they have not dissected what variables influence affect the prevalence of these mood disorders (Skokauskas, Sweeny, Meehan & Gallagher, 2012; Mazaheri et al., 2013).

Coping strategies are amongst other parent factors affecting parental stress, anxiety and depression levels, and may play a moderating role for the effects of other variables. As expected, when parents employ rational, active coping strategies, it is significantly associated with lower levels of stress and depression. However, while there was a visible general trend for anxiety,

with higher levels of rational coping being associated with less anxiety, it was not a significant relationship. When parents who more frequently used rational, active coping strategies were compared to parents who did not utilize these strategies as much, it was demonstrated that the best psychosocial outcomes occurred for the parents with higher rational coping scores. The group with more rational coping skills had significantly less stress and depression than the group with less rational coping abilities. There was still no significant relationship with anxiety. When parents are rational, and better able to figure out what they need to do to stay calm, their ability to remain in control, through implementing these constructive coping strategies, might be able to moderate their stress and depression levels. A key component of stress for these parents results from feeling helpless and experiencing an externalized locus of control in which they do not have control over their situation and outcomes (Lanfranchi & Vianello 2012). When dealing with an above-average number of stressors, employing these rational, active strategies might allow these parents not to become overwhelmed and report as much stress, which could allow them to stay focused and see the end goal, which would also mitigate the risk of depression.

As expected, when parents utilized more avoidant, helplessness coping strategies, then they exhibited significantly higher levels of stress, depression and anxiety. These coping strategies were most strongly associated with higher depression scores, but negatively affected all three components of parental psychosocial well-being. Comparing the parents in the study who utilized more avoidant coping strategies to the parents who these avoidant coping strategies less, there were very large, significant differences between the two groups for both stress and depression; these parents were much more likely to have higher reported levels of stress and depression. While there was a general trend for anxiety, with more avoidant coping strategies being associated with higher anxiety scores, these findings were not significant. Overall,

avoidant coping is not a good sign for a parent's long-term ability to adapt. As explained in the categorization of the coping style, these strategies promote helplessness, which is associated with an externalized locus of control and increased stress levels for parents with children with PWS (Lanfranchi & Vianello 2012). This state of helplessness, compounded with other internal and external factors, is then associated with increased stress, depression and anxiety. Previous research, conducted with typically-developing populations, has shown that there is a bidirectional relationship between avoidant coping strategies and depression, and these results demonstrate that this is generalizable to parents with children with PWS (Steeger, Gondoli & Morrissey, 2013). However, the same study also expressed that there was a unidirectional relationship between stress and avoidant coping strategies, with stress as the pre-emptive factor that led to the development of these strategies (Steeger, Gondoli & Morrissey, 2013). While the present study cannot determine causation or directionality, the findings do support the possibility of a bidirectional relationship for stress and avoidant coping. While increased stress might lead to these coping styles, the increased utilization of avoidant strategies might then augment stress, creating a destructive, cyclic relationship. This presents an opportunity for future exploration for parents with children with PWS, because it could present a focused opportunity for future intervention studies.

After separately examining the effects of child factors and coping strategies, the researchers then explored how different coping methods might be correlated with two important child factors: hyperphagia and severity of behavioral issues. First, looking at rational, active coping strategies, the researchers were surprised to find no significant correlation between these coping methods and either hyperphagia or behavioral problems. However, for avoidant, helplessness coping, there were moderate, significant relationships for both hyperphagia and

behavioral issues. The correlations showed that the more severe either of these child factors, the more the parent utilized avoidant coping strategies. To further explore the compounded effects of children's behavioral issues and coping strategies on parental stress, the researchers then examined the between-group differences in reported stress based on divisions of high or low CBCL scores and coping strategy scores. For both rational, active coping and avoidant, helplessness coping, there were significant between-group differences based on low or high usage of coping strategies, as well as between-group differences based on low or high severity of behavioral issues. For rational coping, the parents reported the least stress when they themselves had a higher rational coping score and their child exhibited less severe behavioral issues. However, based on the average parental stress scores, parent's having lower avoidant coping scores and a child with fewer behavioral issues was the most impactful for decreased stress levels; this was the most desirable combination for the lowest parental stress levels. This shows that while the presence of rational, active coping strategies is beneficial for parents for positive psychosocial outcomes, even more important is the lack of avoidant, helplessness coping strategies. This further reiterates the finding that avoidant coping strategies are more closely associated with negative outcomes for parental anxiety and depression than rational coping strategies are associated with positive outcomes. Therefore, it might be beneficial for future interventions for parents with a child with PWS to first emphasize decreasing the usage of these avoidant coping strategies before then moving on to achieving the usage of positive coping methods; halting avoidant coping strategies should be the initial focus.

The results also demonstrate that although parents sometimes feel like they have no control over their life situations, they can control their coping strategies, and regardless of the children's behavioral issues, the parents who employed more rational strategies and fewer

avoidant strategies exhibited lower stress scores. If parents were made more aware of the concrete nature of the powerful effects of their coping strategies on their own psychosocial outcomes, they might be more open to interventions and also feel as though they were able to exert more control over their own life situations, and feel less helpless. In the end, restoring this sense of autonomy and internal locus of control could be a crucial realization for the success of any intervention studies (Lanfranchi & Vianello 2012).

Finally, surprisingly there were not significant interaction effects on parental stress levels between either behavioral issues and rational coping strategies or behavioral issues and avoidant coping strategies. This demonstrates that there must be some other moderating factor that impacts stress levels, which in turn could have implications for factors affecting anxiety and depression as well. Because of the demographic breakdown of the sample, it further strengthens the idea that there are other factors besides marital status, number of other children and household income that affect these parental psychosocial outcomes. This could be the result of several other external, environmental factors, such as quality of marriage or daily living environment. However, the researchers also propose that another possible moderating variable could be innate parental personality traits.

In the original discussion of the COPE questionnaire, the authors discussed two potential limitations: the potential disconnect someone might experience when their familiar coping strategy is not available to them in a specific situation (e.g. Someone likes to talk to a friend to deal with stress, but does not have the ability to do so if they are temporarily isolated) and the interaction with innate personality traits (Carver, Scheier, & Weintraub, 1989). Therefore, future studies should explore the effects of parental personality traits on coping strategies, as a possible additional moderating factor that could influence the effects of stress on parental depression and

anxiety. For example, there is research indicating that there are differences in coping mechanisms in response to inter-personal family stress, studied through self-reported stress and conflict in step-families, for individuals based on their differential scores for the various Big 5 personality traits (Lee-Baggley, Preece & DeLongis, 2005). There were complex interactions noted between coping mechanisms as well as between all of the various personality traits. Higher scores for specific traits were correlated with differences in coping strategies with some being more maladaptive than others; people scoring higher in neuroticism were less flexible in their coping and generally coped in less constructive ways (Lee-Baggley, Preece & DeLongis, 2005). As well, in a study looking at perceived stress levels in patients with a chronic uncontrollable illness, as a function of their personality scores in each of the five domains of the Big 5 Personality test, neuroticism was a predictor of increase perceived stress (Hasel, Besharat, Abdolhoseini, Nasab & Niknam, 2013). The researchers espoused how “interventions that enhance individual protective factors may be beneficial in reducing stress,” supporting the practical benefits of considering individual personality differences in the development of programs to help populations dealing with chronic stressors manage their high stress levels (Hasel, Besharat, Abdolhoseini, Nasab & Niknam, 2013).

There are several limitations to the study that resulted from the questionnaires used to evaluate some of the variables that provide room for further exploration in future studies. The date from the COPE questionnaire was manipulated in a manner outside of its intended usage, which should be taken into consideration. While the COPE questionnaire has historically been utilized by researchers to assess coping strategies for mothers of children with IDD, the measure was never intended to produce an overall score for coping abilities, and was not meant to determine if a given individual demonstrated one dominant coping strategy (Carver, 2007).

However, for this data analysis the researchers created “High” and “Low” groups for both rational and avoidant coping strategies, in order to employ between group comparisons and demonstrate the differences in outcomes for people utilizing one coping style more strongly than the other. Other coping measures, that do indicate a dominant coping strategy for parents and would be better suited for between-group comparisons, should be considered for future research studies.

Additionally, in the future, other measures of anxiety might be beneficial because the BAI focuses strongly on the serious physical manifestations of anxiety, capturing the severity of the physical presentation of anxiety well but potentially missing key components of what anxiety might look like for parents of children with PWS. Furthermore, the study sample maximum score was only 43, when the highest possible maximum score is 63; the sample is not indicative of the full range of scores for the BAI. Future studies could potentially utilize a different assessment that captures a more complete image of the effects of anxiety, including more questions about the mental state that accompanies the mood disorder. As a result, for this study, the findings related to stress, as measured by the PSI, might be more indicative of the overall anxiousness these parents are experiencing.

In conclusion, this study found that parental stress levels are strongly impacted by their children’s hyperphagic behaviors and the severity of their overall behavioral issues. There were not significant between-group effects for the different genetic subtypes, potentially due to the usage of measures that were unable to capture key components of PWS. Overall, the psychosocial outcomes for parents are strongly affected by their child’s behavioral problems. As well, parental anxiety and depression levels are both strongly associated with parental stress and the severity of their child’s behavioral issues. Parental usage of rational, active coping strategies

is strongly linked to lower stress and depression levels, while parents who employ avoidant, helplessness coping strategies are more likely to have higher levels of stress, depression and anxiety. While there were no significant interaction effects between parental coping strategies and the severity of the child's behavioral issues on parental stress levels, the best outcomes were achieved when parents did not use avoidant coping strategies and their children had fewer behavioral problems. These findings have important implications for improving the baseline understanding of what factors, both parent and child, affect parental mental health and psychosocial outcomes. Future studies have the opportunity to expand upon these findings, and exploring the effects of innate parental personality traits could potentially lead to the discovery of more factors with moderating effects on parental stress. Finally, these findings should serve to increase parental hope and sense of control, because their own coping strategies and personal factors can impact their stress levels. Learning how to develop more effective coping strategies and becoming more cognizant of their own, active role in their psychosocial wellbeing can increase parental sense of control and lower their stress levels, which in turn is associated with a better quality of life and lower levels of depression. This could have important implications not only for future research and interventions, but also for individuals living with PWS and their families.

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Table 1: *Summary of Demographic Data for 154 Participants*

Genetic Subtype		
	<i>Deletion</i>	64.9%
	<i>mUPD (Maternal Uniparental Disomy)</i>	35.1%
Child Gender		
	<i>Male</i>	44.2%
	<i>Female</i>	55.8%
Child Race		
	<i>American Indian</i>	1.9%
	<i>Asian</i>	1.3%
	<i>African-American</i>	5.2%
	<i>Hispanic</i>	3.2%
	<i>Native Hawaiian/Pacific Islander</i>	0.65%
	<i>Southeast Asian</i>	1.9%
	<i>Caucasian</i>	86.3%
Number of Other Children		
	<i>One</i>	40.9%
	<i>Two</i>	25.3%
	<i>Three</i>	14.9%
	<i>4 or More</i>	8.4%
Marital Status		
	<i>Married</i>	75.3%
	<i>Unmarried</i>	24.7%
Household Income		
	<i>Failure to Report</i>	0.065%
	<i>Less than \$15,000</i>	1.3%
	<i>\$15,000 - \$29,000</i>	9.1%
	<i>\$30,000 - \$49,000</i>	11.7%
	<i>\$50,000 - \$69,000</i>	16.2%
	<i>\$70,000 - \$99,000</i>	18.2%
	<i>More than \$100,000</i>	42.9%

Table 2: *Correlations Between Stress, CBCL and Hyperphagia for 154 Participants*

CBCL Total	Pearson Correlation	1	.456**	.637**
	Sig. (2-tailed)		.000	.000
Hyperphagia	Pearson Correlation	.456**	1	.404**
	Sig. (2-tailed)	.000		.000
PSI Total	Pearson Correlation	.637**	.404**	1
	Sig. (2-tailed)	.000	.000	

**Correlation is significant at the 0.01 level (2-tailed).

Table 3: Genetic Subtype’s Effect on Parental Stress and Behavioral Issues for 154 Participants

Descriptives						
		N	Mean	Std. Deviation	Min	Max
PSI Total	Deletion	100	85.570	21.2309	2.1231	37.0
	mUPD	54	91.611	20.1929	2.7479	52.0
	Total	154	87.688	21.0065	1.6928	37.0
CBCL Total	Deletion	100	62.560	9.3033	36.0	94.0
	mUPD	54	65.130	8.3215	39.0	81.0
	Total	154	63.461	9.0280	36.0	94.0

ANOVA				
		df	F	Sig.
PSI Total	Between Groups	1	2.937	.089
CBCL Total	Between Groups	1	2.876	.092

Table 4: *Linear Regression Analysis with CBCL as IV for 154 Participants*

<i>Dependent Measure</i>	<i>R</i>	<i>R²</i>	<i>B</i>	<i>Significance</i>
Stress (PSI) Total	.637	.406	1.483	.000
Anxiety (BAI)	.390	.152	.242	.000
Depression (BDI)	.414	.172	.392	.000

Table 5: *Linear Regression Analysis with PSI as IV for 154 Participants*

<i>Dependent Measure</i>	<i>R</i>	<i>R²</i>	<i>B</i>	<i>Significance</i>
Anxiety (BAI)	.594	.353	.242	.000
Depression (BDI)	.458	.210	.122	.000

Table 6: *Linear Regression Analysis with Rational/Active Coping as IV for 154 Participants*

<i>Dependent Measure</i>	<i>R</i>	<i>R²</i>	<i>B</i>	<i>Significance</i>
Anxiety (BAI)	-.063	.004	-.045	.436
Depression (BDI)	-.294	.086	-.318	.000
Stress (PSI)	-.269	.072	-.715	.001

Table 7: *Linear Regression Analysis with Avoidance/Helplessness Coping as IV for 154 Participants*

<i>Dependent Measure</i>	<i>R</i>	<i>R²</i>	<i>B</i>	<i>Significance</i>
Anxiety (BAI)	.218	.048	.326	.007
Depression (BDI)	.472	.223	1.072	.000
Stress (PSI)	.440	.193	2.455	.000

Table 8: *High and Low Rational/Active Coping- Between Groups Comparisons for 154 Participants*

		N	Mean	Std. Deviation	Min	Max
PSI Total	Low Rational	114	89.912	20.9147	49.0	145.0
	High Rational	40	81.350	20.2010	37.0	120.0
	Total	154	87.688	21.0065	37.0	145.0
BAI	Low Rational	114	5.570	5.9911	.0	43.0
	High Rational	40	4.175	4.2359	.0	14.0
	Total	154	5.208	5.6089	.0	43.0
BDI	Low Rational	114	9.943	9.0211	.0	52.3
	High Rational	40	5.780	6.1503	.0	27.0
	Total	154	8.862	8.5499	.0	52.3

ANOVA				
		df	F	Sig.
PSI Total	Between Groups	1	5.050	.026
BAI	Between Groups	1	1.842	.177
BDI	Between Groups	1	7.309	.008

Table 9: High and Low Avoidance/Helplessness Coping- Between Groups Comparisons for 154 Participants

		N	Mean	Std. Deviation	Min	Max
PSI Total	Low Avoid	125	84.264	20.1735	37.0	133.0
	High Avoid	29	102.448	18.1926	74.0	145.0
	Total	154	87.688	21.0065	37.0	145.0
BAI	Low Avoid	125	4.840	4.5850	.0	18.0
	High Avoid	29	6.793	8.6947	.0	43.0
	Total	154	5.208	5.6089	.0	43.0
BDI	Low Avoid	125	7.396	6.9215	.0	27.2
	High Avoid	125	15.179	11.6772	2.0	52.3
	Total	29	8.862	8.5499	.0	52.3

ANOVA				
		df	F	Sig.
PSI Total	Between Groups	1	19.807	.000
BAI	Between Groups	1	2.889	.091
BDI	Between Groups	1	22.212	.000

Table 10: *Correlations of Rational/Active Coping Strategies and CBCL/Hyperphagia for 154 Participants*

Correlations				
		CBCL	Hyperphagia	Rational, Active Coping
CBCL Total	Pearson Correlation	1	.456**	-.134
	Sig. (2-tailed)		.000	.098
Hyperphagia	Pearson Correlation	.456**	1	.053
	Sig. (2-tailed)	.000		.519
Rational/Active Coping	Pearson Correlation	-.134	.053	1
	Sig. (2-tailed)	.098	.519	

**Correlation is significant at the 0.01 level (2-tailed).

Table 11: *Correlations of Avoidant/Helpless Coping Strategies and CBCL/Hyperphagia for 154 Participants*

Correlations				
		CBCL	Hyperphagia	Avoidance, Helplessness
CBCL Total	Pearson Correlation	1	.456**	.355**
	Sig. (2-tailed)		.000	.000
Hyperphagia	Pearson Correlation	.456**	1	.352**
	Sig. (2-tailed)	.000		.000
Avoidance, Helplessness	Pearson Correlation	.355**	.352**	1
	Sig. (2-tailed)	.000	.000	

**Correlation is significant at the 0.01 level (2-tailed).

Table 12: *Univariate Analysis of Variance for High and Low CBCL and Rational/Active Groups Hyperphagia for 154 Participants*

Between-Subjects Factors					
	Value Label	N	Mean	Min	Max
CBCL Groups	Low CBCL	77	55.618	36	63
	High CBCL	77	69.029	65	75
Rational Groups	Low Rational	114	52.307	36	61
	High Rational	40	63.375	62	72

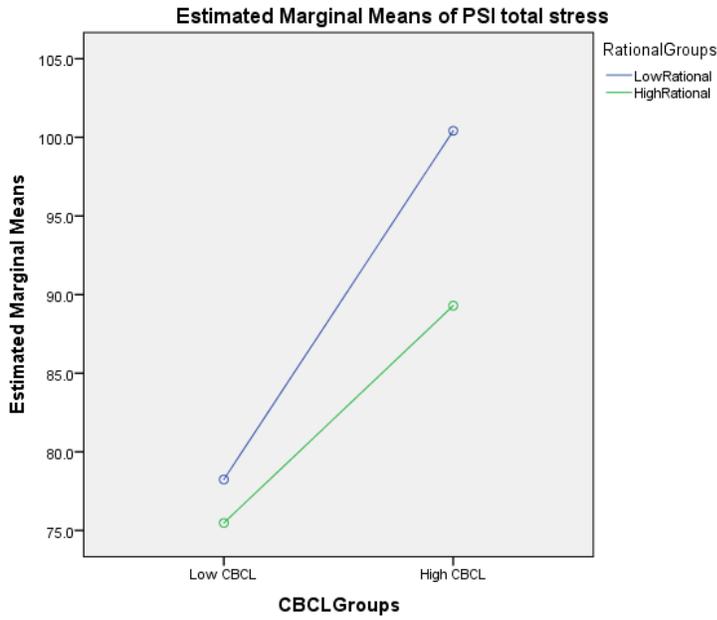
Tests of Between-Subjects Effects			
Dependent Variable: PSI total stress			
Source	df	F	Sig.
CBCL Groups	1	28.551	.000
Rational Groups	1	4.249	.041
CBCL Groups * Rational Groups	1	1.540	.216

Table 13: *Univariate Analysis of Variance for High and Low CBCL and Avoidance/Helplessness Groups Hyperphagia for 154 Participants*

Between-Subjects Factors					
	Value Label	N	Mean	Min	Max
CBCL Groups	Low CBCL	77	55.618	36	63
	High CBCL	77	69.029	65	75
Avoidance Groups	Low Avoidance	125	16.080	12	20
	High Avoidance	29	23.759	21	31

Tests of Between-Subjects Effects			
Dependent Variable: PSI total stress			
Source	df	F	Sig.
CBCL Groups	1	19.325	.000
Avoidance Groups	1	13.984	.000
CBCL Groups * Avoidance Groups	1	.334	.564

Graph 1: *Univariate Analysis of Variance for High and Low CBCL and Rational/Active Groups Hyperphagia for 154 Participants*



Graph 2: *Univariate Analysis of Variance for High and Low CBCL and Avoidance/Helplessness Groups Hyperphagia for 154 Participants*

