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Short term change in psychological well-being of mothers of girls and women with
Rett syndrome

Running head – Mothers and Rett Syndrome

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Abstract

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Keywords: Rett syndrome, mothers, maternal depression, Developmental Behaviour

Checklist, Rett Syndrome Behaviour Questionnaire, longitudinal design

Rett syndrome (RTT) is a rare genetic disorder most commonly caused by a mutation in the methyl-CpG binding protein-2 (*MECP2*) gene, located on the X chromosome at *Xq28* (Amir, Van den Veyver, Wan, Tran, Francke et al., 1999), although RTT remains a clinical rather than a molecular diagnosis (Neul, Kaufmann, Glaze, Christodoulou, Clarke et al., 2010). RTT is associated with severe to profound intellectual disability and a range of other impairments including social and communication limitations (Mount, Charman, Hastings, Reilly & Cass, 2003), developmental regression/loss of skills, motor difficulties, and stereotyped hand movements (Neul et al., 2010). Given this array of difficulties, likely leading to challenges for carers, it is not surprising that researchers have shown increased stress and mental health problems in parents (especially mothers) of individuals with RTT compared to normative samples of adults (Byiers, Tervo, Feyma & Symons, 2014; Cianfaglione, Hastings, Felce, Clarke & Kerr, in press; Laurvick, Msall, Silburn, Bower, De Klerk et al., 2006; Perry, Sarlo-Mcgarvey & Factor, 1992; Urbanowicz, Downs, Bebbington, Jacoby, Girdler et al., 2011), and some indication of increased relationship problems compared to parents of children with other disabilities (Lederman, Alves, Maria, Schwartzman, D'Antino et al., 2015).

Not all parents of individuals with RTT report increased levels of psychological distress. For example, Cianfaglione et al. (in press) found that 24.1% of mothers of women and girls with RTT reported symptoms of anxiety at levels above a clinical threshold, and 5.7% reported symptoms of depression at elevated levels. To understand families that may be at higher risk for psychological problems so that they can be identified and supported, it is important to examine the factors that explain some of the variance in maternal outcomes. Correlates of psychological distress in mothers of individuals with RTT have included: dimensions of the RTT behavioural

phenotype such as fewer physical health complications, fewer stereotypies, and fewer breathing problems (Laurvick et al., 2006); the financial burden of specialist equipment and use of respite care (Urbanowicz et al., 2011), and comorbid epilepsy and parents' beliefs that the individual with RTT may be in pain (Byiers et al., 2014). In a cross-sectional survey, Cianfaglione et al. (in press) found that the severity of the overall RTT behavioural phenotype, assessed using the Rett Syndrome Behaviour Questionnaire (Mount, Charman, Hastings, Reilly & Cass, 2002), predicted increased maternal stress and anxiety. Contrary to general intellectual disability family research findings (e.g., Totsika, Hastings, Emerson, Lancaster & Berridge, 2011) the RTT behavioural phenotype severity, rather than the individual's behaviour problems, predicted maternal psychological distress.

Existing research on the correlates of psychological distress or well-being in parents of individuals with RTT has adopted almost exclusively cross-sectional designs (Byiers et al., 2014; Cianfaglione et al., in press; Laurvick et al., 2006; Perry et al., 1992), and other RTT family research has used retrospective reporting methods (Lederman et al., 2015). In the only study to include data gathered at more than one time point (Urbanowicz et al., 2011), use of specialised equipment and respite care in 2004 were related to maternal mental and physical health two years later. However, these analyses did not account for earlier levels of maternal well-being. Thus, the associations found are difficult to interpret. Prospective research designs are crucial if correlates of maternal well-being are to be clearly understood as risk factors that could be targeted for change in intervention studies.

The purpose of the present study was to use a short term longitudinal follow-up of the Cianfaglione et al. (in press) sample of mothers of girls and women with RTT to examine maternal well-being over time. Our focus was to examine the key

finding from our cross-sectional analysis, but over time. Specifically, we explored whether behavioural and emotional problems or the severity of the RTT behavioural phenotype predicted maternal well-being longitudinally. Our secondary aim was to examine the stability of dimensions of psychological well-being in mothers of women and girls with RTT over a follow-up period of 16-17 months.

Method

Participants

The present study focused on 50 families from which mothers responded to the invitation to participate in a follow-up study of the original survey (see Procedure). These 50 mothers were mainly the biological mother of their daughter with RTT (n=47), with two foster mothers and one adoptive mother. The mothers were 37-70 years of age (mean 51.94 years). Most mothers were living with a partner (n=41), 38% were educated at least to degree level, and 44.4% had family income of under £25,000 (roughly \$40,000 USD) per annum. For 9 families their daughter lived outside of the family home. Their daughters' ages ranged from 5 to 47 years with a mean of 21.04 years: 20 of the individuals with RTT were children and 30 adults at the time of the follow-up.

Measures

Mothers completed three measures about their own well-being. Maternal mental health was assessed with the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983). The HADS was originally developed to allow a quick measure of depression and generalized anxiety in hospital settings, but has been widely used in outpatient and community research. It has been used successfully to measure depression in parents of children with developmental disabilities and maintains good reliability with this population (e.g., Jones et al., 2014). Seven of the

HADS items assess depression (e.g., “I feel as if I am slowed down”), and seven measure anxiety (e.g., “I get sudden feelings of panic”) and each is rated on a four point scale: *most of the time, a lot of the time, from time to time, or not at all*.

The Positive Gain Scale (PGS: Pit-ten Cate, 2003) was used to assess positive experiences associated with raising a child with RTT. The measure consists of seven items; five relating to the perceived benefits for the mother of raising a child with disability (e.g., “Since having this child I have a greater understanding of other people”), and two focusing on what the family has gained (e.g., “Since having this child, my family has become more tolerant and accepting”). Preliminary research findings indicated that the PGS has face and content validity and a Cronbach’s Alpha coefficient of .79 for parents of children with hydrocephalus and spina bifida (Pit-ten Cate, 2003). The PGS has also retained excellent reliability in other samples of parents of children with developmental disabilities (e.g., Jones et al., 2014).

Stressful experiences of raising a child with RTT were measured using the Parent and Family Problems sub-scale of the Questionnaire on Resources and Stress Friedrich-Short Form (QRS-F: Friedrich, Greenberg, & Crnic, 1983). This scale contains 20 items assessing impact on the parent and family (e.g., “Other members of the family have to do without things because of N”, and “N is able to fit into the family social group”). Parents are asked to indicate whether the items are true or false as far as they and their family are concerned. A total stress score is derived by summing the number of negatively endorsed items (i.e., positively worded items are reverse scored). Five items that have been shown to constitute a robust measure of depression in parents of children with disabilities (Glidden & Floyd, 1997) were removed from the scale. This ensured that there was no overlap between the measures of stress and depression in the present research.

Mothers also completed two measures of the behavioural characteristics of their daughter. The *Rett Syndrome Behaviour Questionnaire* (RSBQ, Mount et al., 2002) was used to assess overall severity of the RTT behavioral phenotype. The RSBQ is a 45-item checklist developed to assess behavioural phenotypic characteristics of RTT. Items are rated 0 to 2, where 0 indicates that the item does not apply to the person with RTT, 1 sometimes true, and 2 often true. High internal consistency has been reported for the RSBQ total score (>0.90), with good inter-rater and test-retest reliability scores (>0.80) (Mount et al., 2002).

The extent of behavioural and emotional problems exhibited by each individual with RTT was measured using the Parent Report version of the Developmental Behaviour Checklist (DBC - Einfeld & Tonge, 2002) for children or for adults depending on the age of the individual with RTT. The DBC has excellent psychometric properties (Einfeld & Tonge, 1995; Hastings, Brown, Mount, & Cormack, 2001). The DBC Total Behaviour Score was used as an index of the severity of behavioural and emotional problems displayed by the girls and women with RTT.

Procedure

The survey methodology is described in greater detail in Cianfaglione, Clarke, Kerr, Hastings, Oliver et al. (2015), and the procedure for gathering data from mothers about their psychological well-being is described in full in Cianfaglione et al. (in press). Families were recruited through the British Isles Rett Syndrome Survey (BIRSS), an on-going database currently maintained by the fourth author, with a total of 87 mothers providing data that were reported by Cianfaglione et al. (in press). All of these original 87 families were sent a written invitation to participate in a follow-up study approximately 1.5 years after their initial participation. After reminders, 50

mothers responded (57% response rate). Using information on the dates mothers recorded for the completion of the postal questionnaire survey, the follow-up period was on average 16.44 months (range 15—21 months).

Statistical analysis

To ascertain any pattern of bias affecting the follow-up sample, we compared the 50 follow-up families to the remaining 37 original families on all baseline demographic variables and the five measures used in the current report as scored at the first time point. There were no statistically significant differences between the follow-up sample and the sample who did not participate in the follow-up.

Our secondary research aim was examined by comparing maternal well-being (stress, anxiety, depression, positive gain) between the two time points using paired samples t-tests, and generating stability coefficients for each of these measures over the 16-17 month period.

To address our primary research aim, we built four multiple regression models (one for each maternal well-being measure as reported at follow-up). In each regression model, the total RSBQ score and the total DBC score from the original data collection point were entered as predictors. In addition, the Time 1 score of the relevant maternal well-being measure was entered as a predictor. In this way, any association over time between RSBQ or DBC scores and maternal well-being was examined over and above the stability in the measure of well-being.

Scoring rules were followed for each individual measure in the case of small numbers of missing items from any questionnaire – mean replacement was used where a minority of items were missing from a scale. Where scores were still missing, participants were excluded listwise from analyses involving scores on a missing measure.

Results

Stability of maternal well-being

The mean scores for each maternal well-being measure at both time points, the results of a paired samples t-test comparison, and the 16-17 month stability of the scores (as a correlation r) are displayed in Table 1. Each of the well-being dimensions demonstrated considerable stability both in terms of lack of statistically significant mean change over time and stability coefficients. General mental health scores were more stable than well-being measures that are child-focused (i.e., stress associated with caring for the child, perceptions of positive gain associated with raising the child). However, even the marginally significant mean level change in positive gain scores represented only a very small difference.

-----Insert Table 1 about here-----

Longitudinal regression analyses

The results of the regression analyses are displayed in Table 2. In each model, as expected given the stability of maternal well-being over time, the Time 1 score of each measure was a significant predictor of follow-up scores. Contrary to the results of the cross-sectional survey (Cianfaglione et al., in press), the severity of the Rett syndrome behavioural phenotype at Time 1 (RSBQ total score) was not a significant predictor of maternal well-being. Instead, the overall severity of behavioural and emotion problems (Time 1 DBC total score) emerged as a significant independent predictor of later maternal anxiety and depression scores. The DBC total score did not emerge as a predictor of later maternal stress or mothers' perceptions of positive gain.

-----Insert Table 2 about here-----

Discussion

As with parents of children and adults with disabilities in general, mothers of individuals with RTT experienced stability in stress and mental health problems over a period of 16-17 months. These data suggest that any elevated psychological distress in mothers of individuals with RTT (cf. Byiers et al., 2014; Cianfaglione et al., in press; Laurvick et al., 2006; Perry et al., 1992; Urbanowicz et al., 2011) is chronic – persisting over time. We also reported unique data on short term longitudinal change in the perceptions of positive gain held by mothers of individuals with RTT. Again, these positive perceptions were also relatively stable across 16-17 months. These data are intriguing in that they also suggest that positive perceptions persist over time.

Results from prospective studies in the field of developmental disabilities, across the lifespan, have shown with a degree of consistency that increased frequency or severity of behavioural and emotional problems predicts later parental psychological well-being (e.g., Hartley, Barker, Baker, Seltzer & Greenberg, 2012; Hastings, Daley, Burns & Beck, 2006; Herring, Gray, Taffe, Tonge, Sweeney et al., 2006; Lecavalier, Leone & Wiltz, 2006). Our longitudinal analyses confirmed a similar pattern for mothers of individuals with RTT, with earlier behavioural and emotional problems predicting later maternal anxiety and depression over and above the putative contribution of the severity of the RTT behavioural phenotype. The severity of the RTT behavioural phenotype failed to emerge as a predictor of maternal well-being over time, in contrast to results from our earlier cross-sectional analyses (Cianfaglione et al., in press). This pattern of findings requires replication in additional research, but there is a clear implication that drawing strong conclusions from cross-sectional family research may be misleading.

In addition to the caution that the findings require replication, there are a number of design limitations with the present study that should be considered. First,

the sample was small involving 50 mothers of individuals with RTT across a wide age range. Although longitudinal RTT family research is rare, and so even a sample of 50 may be useful, it is unlikely that this sample would be representative of RTT families. Second, mothers provided data on their daughter with RTT and on their own well-being at both time points. Thus, source variance may be a problem and multiple informant methods are needed in RTT family research in future.

Combined with our earlier cross-sectional study (Cianfaglione et al., in press) the present longitudinal analyses suggest that targeting parental (especially maternal) psychological well-being in RTT families should be an important focus for practitioners and clinical services. Although stress and mental health problems may not be as elevated as for parents of children with other disabilities, mothers of individuals with RTT do appear to be more likely to experience psychological problems and these are likely to persist over time. At the same time, it is important that professionals and agencies recognise that mothers of individuals with RTT hold significant levels of positive perceptions that are also likely to show medium term stability. At present, there is a poor understanding of what functions positive perceptions may serve for parents (Hastings & Taunt, 2002) but these experiences should not be neglected by professionals.

A further practical implication relates to the association over time between maternal well-being and their daughter's behavioural and emotional problems. There have been randomised controlled trial evaluations of parenting interventions showing promise in the field of developmental disabilities (e.g., McIntyre, 2008; Whittingham, Sofronoff, Sheffield & Sanders, 2009) although rarely have existing programmes been tested within families with a child with more profound disabilities (as in RTT). Adapting and testing parenting programmes for mothers and fathers of children with

profound and multiple intellectual disabilities would be a fruitful future avenue for researchers. Successfully reducing their daughters' behavioural and emotional problems is likely to positively affect well-being in parents of individuals with RTT.

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Table 1. Stability of well-being for mothers of individuals with Rett syndrome

Well-being measure	T1 Mean (SD)	T2 Mean (SD)	t	p	Stability (r)
QRS Stress	6.70 (3.22)	7.32 (3.38)	1.55	.128	.63
Positive Gain	29.43 (4.50)	28.20 (5.88)	-1.97	.054	.66
HADS Anxiety	7.82 (4.22)	7.98 (4.28)	.44	.663	.82
HADS Depression	4.69 (3.45)	4.82 (3.76)	.41	.687	.78

Table 2. Regression analyses of maternal well-being

Predictor variable	QRS Stress T2		Positive Gain T2		HADS Anxiety T2		HADS Depression T2	
	β	p	β	p	β	p	β	p
RSBQ total score	.052	.723	.214	.117	.103	.342	-.002	.983
DBC Total score	.096	.507	-.010	.943	.255	.016	.263	.029
QRS Stress T1	.568	<.001	--	--	--	--	--	--
Pos. Gain T1	--	--	.651	<.001	--	--	--	--
Anxiety T1	--	--	--	--	.625	<.001	--	--
Depression T1	--	--	--	--	--	--	.644	<.001

QRS Stress $F(3, 44) = 9.15, p < .001, R^2 = .384$

Positive Gain $F(3, 44) = 12.78, p < .001, R^2 = .466$

HADS Anxiety $F(3, 43) = 36.34, p < .001, R^2 = .717$

HADS Depression $F(3, 44) = 26.67, p < .001, R^2 = .645$

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