Volume 1

Research Component: Literature Review and Empirical Paper

Coping and Psychological Wellbeing in the Caregivers of Children with Rare Genetic Disorders

NATALIE ELIZABETH BYRNE

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Summary

This thesis comprises both research and clinical volumes and is submitted in partial fulfilment of the degree of Doctor of Clinical Psychology (ClinPsyD) at the University of Birmingham.

Volume 1

Volume 1 consists of a literature review, an empirical paper and a public domain briefing. The literature review provides a systematic review of the literature pertaining to parental cognitions relating to behaviours of children with intellectual disability. It has been prepared for publication in *Behavioural and Cognitive Psychotherapy.* The empirical paper reports on parental perceptions of their child's genetic syndrome and examines the way in which this impacts upon parental coping style and wellbeing. This paper has been prepared for publication in *Research in Developmental Disabilities.* This is then followed by a public domain briefing document which summarises the papers in accessible format.

Volume 2

Volume 2 is the clinical component consists of 5 clinical practice reports (CPR's). Please note that all names and identifying information have been altered or omitted and only information of importance to the formulations has been included, in order to ensure anonymity and maintain confidentiality.

CPR1 presents two formulations for "Lucy". Lucy is 32 year old woman. Her presentation is consistent with the presence of Generalised Anxiety Disorder (GAD) as her anxiety seemed pervasive and present in a wide range of contexts and is considered within the backdrop of a lengthy history of anxiety. Basic background and demographic information is presented followed by as summary of the assessment method and data. Lucy's difficulty has been formulated from two perspectives; cognitive behavioural and systemic. The strengths and weaknesses of both

formulations when considering Lucy's difficulties are considered, followed by personal reflections on the process of formulating.

CPR 2 is a case study of "William", a 64 year old man referred to an older adults community mental health team; he had been diagnosed with Psychogenic Nonepileptic Seizures (PNES). The assessment process, and selection of appropriate method for this, is discussed. This is followed by a bio-psycho-social formulation of the development of the PNES. A Cognitive Behavioural Therapy (CBT) intervention was selected and is described. The effectiveness of the intervention is discussed within the evaluation section of the report and is completed with the author's reflections on the therapeutic process.

CPR 3 is a service evaluation aimed to examine the existing national and professional body guidelines relating to Services offered to people with Personality Disorder. The evaluation then sought to compare these recommendations against services already being offered by Psychologists working within Recovery Services in a West Midlands NHS Trust. Data were collected through semi-structured interviews. The findings suggested that at the time of writing, Psychologists were currently delivering services in accordance with the existing recommendations. A number of facilitators of, and blocks to, effective service delivery are identified. Finally, personal reflections on the evaluation and its findings are presented.

CPR 4 reports a single case experimental design and describes the case of "Frank", a 55 year old man presenting with challenging behaviour. Background information and the assessment procedure is described followed by the formulation of Frank's case from the bio-psycho-social perspective initially and then from a behavioural perspective. The intervention is described outlining the principles of reinforcement, extinction and differential reinforcement. The design is then described where an AB methodology was employed. The data collected were subjected to visual and statistical analysis, and a justification of analysis method is given. The findings are then discussed in relation

to outcome of the intervention for Frank, statistical significance and my reflections on the process and outcome.

Finally, CPR 5 was assessed in the form of an oral presentation. As such, only the abstract is included here (although a copy of the presentation slides can be found in appendix 6). The presentation was entitled “Introducing Ellie...” Ellie is a 17 year old young lady. The presentation outlines referral, assessment, intervention and outcomes and this is followed by a discussion of therapist and patient reflections on the process.

Dedication

I dedicate this thesis to my wonderful perfect husband Matthew, my loving, nurturing and amazing mother Sylvia and the best big brother in the world, Richard. My family are my world. Without them there would be no point. I am blissfully happy, *because of you*, and I have survived the most difficult challenges that life has sent me, *thanks to you.* Without you, I could never have achieved this.

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by everyone at the Centre during that time. Some of the data collected by the team have been used in the current study.

Importantly, I would like to thank each and every single one of the unique and inspiring clients that I have worked with over the last three years. I cannot thank them enough, without them, this work would not have been possible.

And for helping me get through the trials and tribulations of clinical training, and for becoming my lifelong friends, (soon to be Dr) Carly Spicer and (soon to be Dr) Leanne Sketchley. My world is a brighter and better place because you are in it.

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Chapter 1- Literature Review

Parental Cognitions, Stress and Coping in Parents of Children with Developmental Disabilities:

A Review of the Literature

Abstract

The current review examines the literature published in the last 10 years, in relation to parental cognitions and their relationship to child behaviour in the parents of children with developmental disabilities. Given that it is generally recognised that mental health difficulties are at least in part influenced and maintained by cognitive differences and style, it was important to examine the impact of parental cognitions relating to their child and their caring role, on parental wellbeing. A literature search was carried out using PSYCHINFO, EMBASE, OVID MEDLINE and WEB OF SCIENCE to identify articles relevant to parental locus of control, self-efficacy and attributions of behaviours in people with intellectual disability. The date of the latest paper presented by Hassall and Rose (2005), provided a cut off for selection of articles for the current review which was set at January 2003. The search yielded 15 papers relevant to the review. The review focused upon parental cognitions of children's behaviour in children with a developmental disability. The literature shows that at present attribution, self-efficacy and locus of control are being measured by different researchers in different ways. To date, there is limited understanding of the way that these three aspects of parental cognition relate. Research is varied in terms of the way that parental cognition is conceptualised, aims and outcome measures, and measurement. It is argued that there is a need to develop a multidimensional model of parental cognition that will be able to fully describe parental cognitions and their relations to child behaviour and parental mental health.

Keywords: developmental disability, parents, attribution, self-efficacy and locus of control.

Introduction

It is recognised that caring for a child with a developmental disability has multiple effects on parents. A long held assumption was that caring for children with a developmental disability would lead to parents experiencing wide ranging difficulties, including common mental health problems such as anxiety and depression (DoH, 2008; Foster et al., 2010; Johnstone et al., 2010; Kim et al., 2003, Padeliadu, 1998; Wulffaert et al., 2009;). Whilst there is strong evidence to suggest that parents of children with intellectual disability do experience mental health difficulties as a result of their caring role, contemporary research also suggests that it is common for parents to adapt to meeting the needs of a child with a developmental disability (Hassall and Rose, 2005) and a growing body of evidence explores the positive contributions that a child with a developmental disability will make to a family and the positive feelings experienced by parents (Hastings and Taunt, 2002; Horsley and Oliver, in press). Given these parallel literatures, it is clearly important to establish the factors which predict and explain why some parents adapt successfully and have a positive experience of caring for a child with a developmental disability and those factors related to a poorer outcome for parents. Given that it is generally recognised that mental health difficulties are at least in part influenced and maintained by cognitive differences and style, it might be beneficial to examine the impact of parental cognitions relating to their child and their caring role, on parental wellbeing.

Cognitive Models Applied to Parenting

Models of Parenting Stress and Coping

Research that has examined the ways that parents think about and subsequently respond to their children, has provided valuable insight into the ways in which parental caregivers interpret child related events and how these interpretations are linked to emotional and behavioural responses (Bugental et al., 1998). There are a number of cognitive models which have been applied in this research that were reviewed by Hassall and Rose (2005). Within their review Hassall and Rose discuss different models of parental stress and coping. They include the "Double ABCX" model (McCubbin and Patterson, 1983 cited by Hassall and Rose, 2005), the general model proposed by Mash and Johnstone (1990) and the cognitive theory of stress and coping (Lazarus and Folkman, 1984 cited in Hassall and Rose, 2005). They concluded that each of these models vary in terms of the level of complexity and focus but suggest that they are all based on a multidimensional model. They suggest that both parental cognitions and appraisals are important elements to each of the models and that each of these models, which have received differing levels of empirical support, describe the way in which parental factors (including parental cognition) exert an influence on coping styles and subsequent outcomes. Therefore, within the literature there is a broadly accepted notion of a model which combines parental cognitive factors, stress and coping and other outcomes. Hassall and Rose conclude that no single model that adequately explains and conceptualises these factors and their relationships has been proposed. A decade later it is important to consider studies published since the Hassall and Rose review which seek to explore parental cognitions generally and their relationship to stress, coping and adaptation as these might progress model building in this area.

Attribution Models

One of the most significant contributions to understanding parental cognitions has been attribution theory. According to Bugental et al., (1998), attributions can be understood as a type of "interpretive filter through which meaning is assigned to the behaviours and characteristics of the child and the nature of the parent-child relationship" (pp 460, Bugental et al., 1998). They also suggest that a parent's emotional response tends to reflect the attributions that a parent holds. To understand the attribution literature in relation to parental caregivers, it is first essential to go back to its earliest origins. In 1980 Weiner proposed an attributional model which considered the dimensions of locus, intentionality, stability and controllability as forming a person's situation specific attribution (Weiner, 1980; Weiner, 1985; Bugental et al., 1998). Since its early development, research which has examined attribution theory has used a variety of definitions and measurements (Bugental., 1998). Within the subsequent literature this original model appears to have been separated into three different theoretical frameworks which include attribution, self-efficacy and locus of control. The reasons for this are explained in Hassall and Rose's proposition that attribution tends to be a momentary, spontaneous and moment specific cognition, whereas locus of control and self efficacy represent more schematic cognitive processes. Originally proposed by Rotter (1966), the concept of locus of control refers to the extent to which a person feels that they have control over the events in their lives. It may be reasonable to assume that a person will have varying perceptions of the level of control which they have depending on the situation that they are in, and these will show a degree of consistency based on the features of that situation. Consequently, locus of

control could be considered a schematic cognitive process as suggested by Hassall and Rose (2005).

Attribution theory as a model of understanding child related parental cognitions has received a great deal of attention within the literature. As a result it is beneficial to develop a better understanding of the ways that attribution theory has been applied to parental caregivers and an understanding of how useful an endeavour this has been.

More recently it has been proposed that a multi-dimensional model which incorporates attribution, self-efficacy and locus of control, may be preferable for understanding parental cognitive processes (Hassall and Rose, 2005). There has been no research to date which has examined all of these aspects of "parental cognition" simultaneously. As such is difficult to conclude if attribution, self-efficacy and locus of control are independent concepts or the degree to which they overlap. This might suggest that attribution theory alone is insufficient as an explanation for parental cognitive responses to their child and their child's behaviour and the outcomes of these. It is important to consider more recent literature to determine whether the different aspects of parental cognition continue to be examined independently or if a more coherent picture is emerging.

Attribution Models, Stress and Coping

It is essential to understand parents cognitive responses to their child and their child's behaviour, due to their links to important outcomes such as help seeking behaviour, parenting responses and also parental and child wellbeing. Chavira et al., (2000) found that in mothers of children with developmental disability, those who reported attributing

responsibility of problem behaviours to the child also reported feelings of anger and frustration and more aggressive/harsh behavioural reactions. Further to this, although not investigating the parents of children with developmental disability, Morrisey-Kane and Prinz (1999) reviewed the literature that linked parental attributions and expectations and considered how these related to help seeking behaviour, engagement and retention in treatment and outcome, which is particularly relevant for parents of children with developmental disability. They concluded that parental attributions at the point of commencement in treatment, can predict likelihood of attrition. Therefore, those at risk could be identified at first point of contact within the treatment setting (Morrisey-Kane and Prinz, 1999). They go on to suggest that future research should focus on the development of models and measures that will be better at capturing the complex interaction between child behaviour, parenting cognitions and parenting behaviour. This would indicate that Morrisey-Kane and Prinz (1999) believe that Weiner's (1980) attributional model is not entirely sufficient for describing parental cognitions and their association to emotional responses and parental behaviour.

There is now agreement that parental wellbeing is affected by a number of factors which include characteristics of the child, family and environment and specifically parental cognitive responses to their child and their child's behaviour (Hassall and Rose, 2005). It is argued that these factors form part of a multi-dimensional model of stress and coping in parents of children with developmental disability. However, Hassall and Rose (2005) state that despite the fact that parental cognitive responses, stress and coping are necessarily related, there seems to be little overlap between the literatures in both of these areas when in

The Hassall and Rose Review (2005)

Due to the wide variation in studies, a review would aid understanding of the cognitive factors which influence parents' responses to their child and their child's behaviour and parental mental health and coping. Such a review was conducted by Hassall and Rose (2005). They began with a conceptual review of the literature which sought to provide an overview of the findings in relation to parental cognitions and stress in the parents of children with a developmental disability. Based on this, they reviewed the literature systematically examining the three cognitive variables relevant to the study of stress and coping in parents of children with developmental disability: parental attributions, self-efficacy and locus of control. Given the limited number of articles that were specific to parents of children with developmental disability, Hassall and Rose (2005) also reviewed the literature pertaining to parental cognitions for children with various difficulties and included those relevant to intellectual disability.

Parental Attributions

When considering parental attributions, Hassall and Rose (2005) concluded that the way that attributions are conceptualised in the literature varies and as a consequence, stated that it is difficult to draw firm conclusions about the impact of various forms of parental attributions on outcomes. However, they established a degree of consistency in the findings of literature which they considered within their review. They reviewed parental attributions in children in non-clinical samples, in clinical samples and the few papers which focused on parental attributions in parents of children with a developmental disability. Their findings provide a complex picture of attributional patterns. Within the literature examining

parental attributions of positive and negative behaviour in children in a non-clinical sample, parents tended to attribute positive behaviour to internal, innate and stable child characteristics with negative behaviour being attributed to external causes. However it seems that the literature pertaining to parental attributions of behaviour in clinical samples may not be the same and they provide evidence of alternative and distinctive patterns of attributions. For example, in the parents of children with behavioural disorders and those mothers with depressed mood, attributions of problem behaviours tended to be internal to the child rather than externally determined (Hassall and Rose, 2005). In contrast, in the parents of children with ADHD, a more complex pattern of attributions was found. Positive child behaviours tended to be attributed to the child's disposition whilst attributions relating to negative child behaviour in these parents were influenced by whether the child was medicated or not (Hassall and Rose, 2005). They suggested that the attributions made by the parents of children with ADHD may be influenced by beliefs about the origins of the ADHD (either biological or environmental for example) and concluded that this pattern might also be observed in parents of children with intellectual disability considering it is also a chronic condition. This is supported by the findings of Chavira et al., (2000) who found that Latino mothers of children with a developmental disability did not think that their children were responsible for their problem behaviours. They also found that when mothers attributed responsibility for problem behaviours to the child, they were more likely to experience negative emotional outcomes.

To conclude, Hassall and Rose suggest that there is a predictable pattern to parental attributions such that generally internal and stable attributions are made in relation to positive behaviours, whereas external and unstable attributions are made in relation to

negative behaviours within clinical samples. This proposition, they suggest, has received empirical support in the parents of typically developing children and has been replicated for parents of children with a developmental disability (Chavira et al., 2000). Hassall and Rose (2005) concluded however, that this pattern of attributions may not be consistent across all groups of parents and children and that other factors, such as nature of the child's difficulty, might also exert an influence over attributions made. They suggest that this should be considered and incorporated into future research.

Parental Self-Efficacy

As with other aspects under investigation in their review, Hassall and Rose concluded that there are very few published studies which examine the self efficacy in parents of children with a developmental disability. They noted that there is inconsistency in the terms used to describe and examine self-efficacy in the literature and state that "parenting competence" and "parenting self-esteem" seem to be used interchangeably and therefore recommend that these concepts are combined and described as "parenting self-efficacy". They suggest that combining both terms conceptually to examine parenting efficacy might be an area worthy of further exploration.

Parental Locus of Control

In terms of parental locus of control, Hassall and Rose stated that the majority of studies have used a general measure of locus of control rather than using a parenting specific scale such as the *Parenting Locus of control Scale* (*PLOC*; Hassall et al., 2005). When reviewing this literature, evidence suggested that low parenting control (as measured by the *PLOC*) was associated with higher levels of parenting stress and psychological distress in parents of

children with a developmental disability. They suggested that further research exploring parenting locus of control should employ specific rather than general measures of locus of control using a tool such as the *PLOC* as this would allow for comparisons within the literature.

Hassall and Rose (2005) noted that the vast majority of research focuses on mothers rather than fathers. They cite three studies which have found parent gender to have no impact on level of stress experience which might suggest that the findings of the literature focusing solely on mothers can be generalised to all parents. However, they also acknowledge that there may be different aspects of the family environment which cause stress for fathers and not mothers. They also state that there are a number of factors which have been demonstrated consistently to be mediating factors in parental stress and coping and cite a number of review articles which have demonstrated this.

Hassall and Rose found that research continues to examine parental self efficacy, locus of control and attributions independently from the others. At present, it seems that patterns of attributions are complex and influenced by a number of factors. They also found that it was difficult to draw together findings relating to parental locus of control due to differences in measures and the lack of consistent use of a measure designed specifically for use with parents. Based on their recommendations, it is therefore important to consider whether there has been a move forward to examining parental locus of control in these studies rather than employing general measures of locus of control.

The Hassall and Rose paper summarised succinctly the literature relating to parental cognition, stress and coping. They drew a number of theories and concepts together and discussed the way in which these theories can be combined to aid understanding of how parental cognitive processes influence stress and coping in parents of children with a developmental disability. They argue that parenting self esteem and locus of control are general schemas that relate to a person's ability to manage a situation, whereas parental attributions can been seen as being more situation specific. They suggest that in combination, these "parental cognitions" can be seen as a moderator of parental stress, coping and adaptation. They suggest that further investigation would be helpful and would provide valuable information that could influence the development and delivery of clinical interventions for children who may be displaying behavioural problems.

Given the recommendations made by Hassall and Rose, the current review will examine the recent literature in relation to parental cognitions, and their relationship to child behaviour in the parents of children with a developmental disability. Examination of the Hassall and Rose reference list revealed the latest articles within their review were published in 2003. As a result this current review will explore the literature published since 2003 and consider this in light of the Hassall and Rose recommendations and the theoretical links as outlined above. The current review will: 1) examine and describe studies exploring parental cognitions about their child's behaviour and their relationship to stress, coping and adaptation, 2) evaluate whether the different aspects of parental cognition are being examined independently or in combination, 3) examine whether there has there been a move forward to examining parental locus of control in these studies rather than employing general measures of locus of control.

Search Strategy

A literature search was carried out in July 2013 using PSYCHINFO, EMBASE, OVID MEDLINE and WEB OF SCIENCE to identify articles relevant to parental locus of control, self-efficacy and attributions of behaviours in people with a developmental disability. Table 1 lists the search terms that were employed. Since Hassall and Rose conducted a similar review which was published in (2005), the date of the latest paper presented, provided a cut off for selection of articles for the current review which was set at January 2003.

Table 1. Terms used in the literature search for articles describing parental attributions of behaviours in their children with intellectual disabilities.

Search Term	**Variations**
Parents	Parent*, mother*, father*, maternal, paternal
Cognitive	Attribu*, parenting, cognitive, style, cognition*, belief*, locus of control, perception*, self esteem, efficacy
Behaviour	Behavio*
Developmental disability	Intellectual disab*, learning disab*, mental retard*, mentally retarded, mental handicap*, mentally handicap*, developmentally disab*, developmental disab*, developmental difficult*, intellectual impairement*, intellectually impaired, cognitive impair*, autis*, autistic spectrum

* Search included both singular and plural terms e.g. parent and parents

The initial search yielded 2786 articles (once duplicates were removed). The results were then inspected and irrelevant papers were excluded. This resulted in 100 papers of interest; the abstracts of which were then inspected and all papers which did not meet each of the search criteria were removed. Papers were considered irrelevant if, for example, they did not focus on parental caregivers. This yielded a final 14 papers for inclusion in the literature review. Of the 14 papers, all references were inspected to identify any further relevant literature. This yielded one additional paper for inclusion in the review, giving a total of 15 papers.

Exclusion criteria

To ensure that only peer reviewed research was included in the review all book chapters, conference presentations and dissertation abstracts and articles not written in English were excluded. Finally articles were excluded if they did not include each of the criteria of interest; i.e. if they did not consider parental cognitive factors, discuss child behaviour and if the population under examination was not parents of children with a developmental disability.

Results and Discussion

A summary of these papers can be found in Table 2. Details relating to aims of the study, the sample, methodology, measures and outcomes are included.

Table 2. Summary of articles

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Armstrong & Dagnan (2011)	Applies Weiner (1980)'s attributional model to maternal attributions of challenging behaviour. Exploratory study examining maternal attributions, assignment of responsibility and emotional and behavioural responses to the challenging behaviour. Also examines anger as a mediator to behavioural response when an attribution of responsibility is made to the child's challenging behaviour.	Mothers (n = 56) 18.7% response rate	Quantitative Questionnaires in relation to 3 vignettes each describing challenging behaviour	• *Modified Attribution Style Questionnaire* (Peterson et al., 1982) • Anger & Sympathy- 7 point Likert scales • Scale of likelihood of punishment (Graham et al., 2001) • Perception of child's disability- 7 point Likert scale • Perception of child's challenging behaviour- 7 point Likert scale	Behaviour type influences the attribution process When child considered actively engaged in the challenging behaviour, mothers assigned more control and responsibility to the child, experienced more anger and were more likely to punish the behaviour

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Choi and Kovshoff (2013)	Explores the relationship between parental attributions and the acceptability of treatment for problem behaviours in children with autism spectrum disorder.	Mothers (n = 139) Convenience sample so response rate n/a. Authors posted adverts for the study on relevant websites and contacted various organisations (support groups, schools and charities) and sought permission to send out invitations via their mailing lists	Quantitative	• *Parental attributions questionnaire* (*PAQ*; Whittingham et al, 2008, 2009) • Treatment acceptability measure (case vignette, treatment descriptions, and the *treatment evaluation inventory-short from; TEI-SF*- Kelley et al. 1989) • *The conduct problem subscale of the NCBRF-parent version* (Aman et al., 1996; Tasse et al., 1996) • *The Social Communication Questionnaire-Current Versions (SCQ;* Rutter et al., 2003)	When parents perceived parent-related causes for their child's problem behaviour, they were less likely to find parent-focused behavioural interventions acceptable. None of the child-referent attributional dimensions were associated with treatment acceptability of the parent focused-behavioural programmes which is inconsistent with previous research findings. No relationships found between any attributional measures and the child-focused interventions. As severity of child's disruptive behaviour increased, acceptability towards behavioural interventions reduced.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Dale, Jahoda & Knott (2006)	Applies Weiners (1980) attributional model to Mother's of children with ASD. Exploratory study examining the nature and impact of attributions on depression, parenting stress and expectations about the child's future.	Pilot Phase- Mothers and Fathers (n = 9) Main Phase- Mothers (n = 16) 50% response rate	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires) Pilot phase- focus group in order to develop a semi-structured interview. Main phase- Participants were interviewed and completed a number of questionnaire measures.	• *Parenting Stress index* (3rd Ed: Abdin, 1995) • *Child expectations Scale* (Dunst & Trivette, 1986) • *Beck Depression Inventory* (2nd Ed: Beck et al, 1996)	Mother's made a diverse and complex range of attributions about their child and their child's future (related to cause stability, controllability) all of which were related to difficulties associated with autism spectrum disorder such as lack of certainty around cause and prognosis.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Drysdale, Jahoda & Campbell (2009)	Applies Weiners (1980) attributional model to mother's of children with intellectual disability who engage in self injurious behaviour. Exploratory study examining parental attributions of their child's self injurious behaviour and the impact this has on maternal wellbeing and engagement in treatment	Mothers (n =13) Convenience sample so response rate n/a. Sample was taken from a combination of statuatory agencies and voluntary organisations. No further details were provided	Qualitative	Semi-structured interviews including topics of perceived causes of self injurious behaviour, responses to self injurious behaviour and feelings of stress and self efficacy. Semi-structured interview was developed based on the thematic analysis of two interviews with mothers who were asked to recall and reflect upon their experiences of self injurious behaviour in their children.	Attribution consistent with Weiner's (1980) dimensions of cause, stability and controllability. There were different and contradictory views held about the causes of the SIB. Mothers were pessimistic about any long term improvement in the self injurious behaviour and uncomfortable with suggestions made by professionals. Perception of limited control over SIB and control was linked to stress and a sense of responsibility in the mothers who had inadequate support.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Gale (2009)	Examines how locus of control is expressed in caregivers descriptions of children's behaviour	Parents and caregivers (n = 28 transcripts from 15 caregivers- seven parents, five teachers and 3 carers)	Qualitative	Structured interview designed to generate responses that might indicate parental locus of control	Locus of control was situation specific and parents "Locus of Control" was less clearly identifiable than what had previously been observed in the literature based on more standardised forms.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Gore & Umizawa (2011)	Evaluation of a training programme for the family carers (and staff) of children with intellectual disability. Exploratory study examining the impact of the training programme of attributions made by family carers of challenging behaviour prior to and post training. Also measures emotional reactions to the challenging behaviour	Family carers (parents, siblings or grandparents; n = 49)	Quantitative	• *Checklist of Challenging Behaviour (CCB*, Harris et al., 1994) • *Challenging Behaviour Attributions Scale (CHABA*, Hastings, 1997) • *Emotional Reactions to Challenging Behaviour Scale (ERCB*, Jones and Hastings, 2003)	Study findings presented in terms of the whole sample (this included staff working with the children too) Reduction in the severity and frequency of challenging behaviour post training. Whole group were less likely to attribute emotional factors to the challenging behaviour and there was a reduction on the anger and depression subscale post training.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Hassall, Rose & McDonald (2005)	Investigated the relationship between parental cognitions, locus of control, child characteristics, family support and parenting stress for mothers of children with intellectual disability.	Mothers (n = 46) 23% response rate	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires)	• *Vineland Adaptive Behaviour Scales- 2 (VABS*; Sparrow et al., 2005) • *Family support scale (FSS*; Dunst et al., 1984) • *Parenting Sense of Competence Scale (PSOC*; Gibaud-Wallaston & Wandersman 1978, as cited in Johnston and Mash, 1980) • *Parental locus of control - shortened version (PLOC*; Hassall et al., 2005)	Child behaviour difficulties associated with parenting stress. Mothers with greater levels of social support reported lower levels of stress. Parenting stress was associated with behavioural difficulties of child, locus of control and parental satisfaction. This relationship was partly mediated by family support.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Hill & Rose (2009)	Examined locus of control, parenting self esteem and parental cognitions in the mothers of adults with an intellectual disability.	Mothers (n = 44) 20% response rate	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires)	• *Vineland Adaptive Behaviour Scales- 2 (VABS*; Sparrow et al., 2005) • *Family support scale (FSS*; Dunst et al., 1984) • *Parenting Sense of Competence Scale (PSOC*; Gibaud-Wallaston & Wandersman 1978, as cited in Johnston and Mash, 1980) • *Parental locus of control - shortened version (PLOC*; Hassall et al., 2005) • *Parenting Stress Index- short form (PSI-SF*; Abidin, 1990)	Regression analysis revealed a relationship between adaptive behaviour, behavioural difficulties and parenting stress. Mothers with greater levels of social support experienced lower levels of parenting stress. Mothers with a more internal locus of control reported lower levels of parenting stress. Parenting satisfaction mediated the relationship between adaptive behaviour and parenting stress.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Hodgetts, Savage and McConnell (2013)	Investigated the experience and perceived outcomes of the stepping stones triple P programme for the parents of children with autism.	Parents (mothers =8, fathers = 2)	Mixed Design- Multiple case study Qualitative (Interviews) & Quantitative (Questionnaires)	• *Depression- Anxiety-Stress Scale (DASS*; Lovibond and Lovibond, 1995) • *Parenting Self efficacy questionnaire* (Hastings and Symes, 2002) • Supports and Services Questionnaire- designed for the purposes of the current study • Semi-structured interviews to illicit participants stories about the child with autism, family life, adaptive resources and outcomes.	Three key themes emerged from the interviews 1) changes in causal attributions of misbehaviour, 2) "who's the boss?" parents feeling more in charge of their child's behaviour and 3) appreciation of the positive approach to behaviour management.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Lloyd and Hastings (2009)	Investigated the parental locus of control and its role in relation to maternal wellbeing.	Mothers (n = 91)	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires)	• *Vineland Adaptive Behaviour Scales- 2 (VABS*; Sparrow et al., 2005) • *Strengths and Difficulties Questionnaire (SDQ*; Goodman, 1997) • *Questionnaire on resources and stress: Freidrich short form (QRS-F*; Friedrich et al., 1983) • *Parental Locus of Control* (*PLOC*; Campis et al., 1986) • *Positive contributions scale (PCS*; Behr et al., 1992) • *Hospital Anxiety and Depression Scale (HADS*; Zigmond and Snaith, 1983)	External locus of control was associated with maternal distress. Regression analysis showed that maternal positive perceptions could be predicted by parental locus of control; Mothers who believed in fate/chance more likely to appreciate positive aspects of the child, those who felt that they could not control their child's behaviour were less likely to make positive appraisals of the child Locus of control relatively stable over time.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Meirsschaut, Roeyers & Warreyn (2010)	Exploration of parenting experiences of mothers of a child with autism spectrum disorders in compared to parenting their typically developing child	Mothers (n = 17)	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires)	• *Maternal efficacy scale* (Teri and Gelfand, 1991) • *Maternal agency questionnaire (*Kuhn and Carter, 2006) • *Maternal guilt questionnaire* (Kuhn and Carter, 2006) • *Nijmeegse Ouderlijke stress index* (De Brock et al., 1992)	"It affects our whole life"... describes the impact of having a child with autism spectrum disorders. Higher levels of stress related to parenting incompetence and mores symptoms of depression concerning their child with autism spectrum disorders. Felt more guilty about "not doing enough" to stimulate the development of their typically developing child.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Whittingham, Sofronoff and Sheffield (2006)	Main focus of study was to assess acceptability of strategies contained within the Stepping Stones Triple P programme. Additional exploration of the impact of parental attributions and parental perceived control in terms of their acting as potential barriers to positive expectations of the programme.	Parents (n = 42) Focus group (Mothers n = 4) Study (Mothers n = 34, Fathers n = 8)	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires)	• Focus group to generate qualitative data • *Attribution and Control Questionnare (ACQ;* Whittingham et al., 2006) this measure was designed specifically for the current study • *Parenting Strategies Questionnaire* (Whittingham et al., 2006) this measure was designed specifically for the current study • Follow-up questionnaire(Whittingham et al., 2006) this measure was designed specifically for the current study	Responses to programme generally positive. Attribution of child's behaviour to uncontrollable forces found to predict higher ratings of usability of the programme Findings interpreted in the context of Weiner's (1980) model

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Whittingham, Sofronoff, Sheffield & Sanders (2008)	Exploration of parental attributions within the autism spectrum disorders population	Parents (n=59: 54 mothers, 4 fathers, 1 grandmother) Sample the same as Whittingham et al., (2009)	Quantitative	• *Family background questionnaire* (Sanders et al., 2003) • *Autism-Spectrum Quotient (AQ*: Sanders et al., 2003) • *Parental attribution questionnaire (PAQ*: Whittingham et al., 2006)- designed specifically for the purpose of the current study	Parents with greater degree of autistic traits were more likely to believe that they had caused their children's autism.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Whittingham, Sofronoff, Sheffield & Sanders (2009)	Part of wider project- looked at the effect of parental attributions on the effectiveness of a parenting programme (stepping stones) for parents of children with autism spectrum disorders	Parents (n=59: 54 mothers, 4 fathers, 1 grandmother)	Mixed Design Qualitative (Interviews) & Quantitative (Questionnaires)	• Semi-structured interview in preliminary stages to verify diagnosis of autism spectrum disorders • *Family background questionnaire* (Sanders et al., 2003) • *Eyberg child behaviour inventory (ECBI*; Eyberg and Pincus, 1999) • *Parenting scale* (Arnold et al., 1993) • *Parental attribution questionnaire (PAQ*: Whittingham et al., 2006)	Parental attributions altered as a result of the programme. Parents less likely to believe that misbehaviour was intrinsic to the child. Following programme parents were more likely to believe that their child's autism spectrum disorders may change in the future. Parental attributions pre programme predicted change in dysfunctional parenting styles of over-reactivity and verbosity.

Author	Summary of Research	Sample	Methodology	Measures Used	Outcomes
Woolfson, Taylor & Mooney (2010)	The study aimed to explore the role of parental attributions of adult and child controllability as a moderator of the relationship between disability and problem behaviours.	Mothers (n = 46) Opportunity sample so response rate n/a. Advertisements were placed on general websites and websites for special groups.	Quantitative	• *Child behaviour checklist (CBCL*; Achenbach & Rescorla, 2001) • *Parent attribution test*- adapted version for current study (*PAT;* Bugental, 2004)	Authors suggest that the findings reflect existing literature- Developmentally delayed children presented more behavioural difficulties than typically developing children (aggressive behaviour, rule-breaking, social problems and other problems). Effect of group on problem behaviours was moderated by parental attributions of their own controllability. No difference between groups when parents had high adult controllability suggesting that high adult controllability attributions might have a positive impact on the parents of developmentally delayed children.

Review Strategy

Upon inspection of the articles included within the current review, it became clear that, despite the search terms used, the papers did not provide a clear narrative of the area under investigation. Articles were not theoretically or methodologically similar (as will be described in detail below) and consequently a decision was made not to review the articles according to a quality framework, as is commonplace within academic literature reviews. Instead, where themes are identified as either absent or present, observations and summaries will be made. It is argued that there is little value in reviewing the quality of the literature, if the articles are not relating in a way that will advance understanding of the concepts and the ways in which they relate within a given area of research.

Methodological Limitations

Data Measurement and Analysis

Studies examining parental cognitions have used qualitative, quantitative and mixed research methodologies.

The Use of Qualitative Methods

A number of qualitative methods were employed within the articles in this review. Drysdale et al., (2009) provided an account of mother's attributions of self injurious behaviour in the children with a developmental disability and Gale (2009) explored the way in which parents

and caregivers express locus of control in descriptions of their child's behaviour using interview methods. Woolfson et al., (2010) employed semi-structured interviews to explore mothers experience of parenting a child with autism and a typically developing child, they then explored this further using quantitative methods. Qualitative methods were also employed by Dale et al., (2006) who held a focus group to develop a semi-structured interview. Whittingham et al., (2009) employed qualitative methodology in the form of a semi-structured interview for the purpose of verifying the diagnosis of ASD within the sample. Hodgetts et al., (2013) used semi-structured interviews to illicit further qualitative date within the context of a multiple case study design and one study used a focus group to explore further its quantitative findings (Whittingham et al., 2006). A further three studies employed qualitative methods using a semi-structured interview to complete the *Vineland Adaptive Behavior Scale (VABS;* Sparrow et al., cited in Hill and Rose 2009). However, in each of these studies only the quantitative aspects of the *VABS* are reported (Lloyd and Hastings, 2009; Hassall et al., 2005; Hill and Rose, 2009).

All of the qualitative methods employed in the above studies can be described as qualitative self report (Barker et al., 2002). The obvious advantage to such methods is that they allow for exploration of the subject matter and provide access to phenomenological data which could not have been accessed in any other way (Barker et al., 2002). There are however, a number of disadvantages to such methods. For example, they do not produce standardised outcomes, they do not allow for direct comparisons, cannot be replicated and they are also subject to researcher bias (Barker et al., 2002) in terms of the analysis. When a literature is predominated by qualitative methodology, the validity and generalisability of the findings

can be questioned. It is noteworthy that qualitative methodology tends to be employed as a means of gathering data that would allow for the building of a theory or model which is then generally tested using quantitative techniques and this will be discussed further below.

The Use of Quantitative Methods

Questionnaire measures are often employed to allow for the collection of large amounts of data and to provide an opportunity for comparison and replication of the findings to other studies and populations (Barker et al., 2002). The advantage of these quantitative methods is that data can be subjected to analyses which can establish the direction of a relationship if the design is appropriate. However, the quantitative studies under review here are often analysed using correlational analyses (Choi and Kovshoff, 2013; Hassall et al., 2005; Lloyd and Hastings, 2009), which suggests that at this point these papers continue to be theory-model building rather than testing.

Qualitative vs. Quantitative Methods

Qualitative research tends to reflect the immature status of a literature in which there is no clear theory or model developed. Within the current review two of the studies employ purely qualitative methods (Drysdale et al., 2009; Gale, 2009) with a further eight studies employing a mixed methodology. This also suggests that at this point there is still no clear consensus within the literature about parental cognitions and how these are linked to outcome and moderating variables. This reflects the fact that there are a number of different concepts under the overarching idea of "parental cognitions" and also that parental

cognitions are being linked to a wide range of outcomes in the literature, none of which appear to have been fully explored at this point.

Development of New Measures

Within the papers under review there are a number of instances where measures have been developed (Armstrong and Dagnan, 2011; Drysdale et al., 2009; Gale., 2009; Hodgetts et al., 2013; Whittingham et al., 2006; Whittingham et al., 2008). Three of these developed quantitative measures, the other three semi-structured interviews. For those studies which have included newly developed questionnaire measures, when Cronbach alphas have been reported, some of the scales do appear to evidence good internal consistency (Armstrong and Dagnan, 2011). Hodgetts et al., (2013) acknowledge that they did not provide an assessment of consistency for their measure, but state that this would not have been possible given the small number of participants. Finally, within the development of the *Parental Attribution Questionnaire* (*PAQ*; Whittingham et al., 2008), one of the subscales had low internal consistency and whilst this has been acknowledged by the authors, there does not appear to be a clear understanding of why this is the case.

In the same way that there appears to be no clear and consistent conceptualisation of parental cognitions or a model in which to place and understand these, there appears to be a tendency to develop new and unique measures to measure the same construct within different studies and populations. Clearly defining parental cognitions, whether that be a combination of attribution, locus of control or parental self efficacy, or not, would allow for the development of robust measures for use within the developmental disability population.

Sampling Issues

Within the current studies there are a number of issues with regard to sampling methods. There is significant bias within some samples. Of the fifteen studies, all employed a convenience sampling method, for example, sampling from all of the parents with children at particular schools (e.g. Gore and Umizawa, 2011; Hassall et al., 2005; Lloyd and Hastings, 2008), in a particular geographic region (Choi and Kovshoff, 2013), those accessing support groups and/or websites (Dale et al., 2006; Woolfson et al., 2010) or accessing a particular service (e.g. Drysdale et al., 2009; Hill and Rose, 2009; Hodgetts et al., 2013; Meirschautt et al., 2010; Whittingham et al., 2006; Whittingham et al., 2008; Whittingham et al., 2009). There are obvious biases, within this sample of participants. For example, the parents accessing support groups may be those parents who have an active style of coping, and may be experiencing greater or lesser levels of distress as a result of their child's behaviour. Given that some of the parents were those accessing a particular service, the study methodology cannot explain any difference between these parents and their children and those who do not access such services. Finally, some report particularly low response rates (Hill and Rose 2009; Armstrong and Dagnan, 2011) and this must also be taken into consideration when interpreting the findings.

Parents: Mothers or Fathers?

In terms of the sample under investigation there is a bias towards mothers. Nine of the papers reported samples of mothers only (Hassall and Rose, 2005; Hill and Rose, 2009; Meirschautt et al., 2010; Dale et al., 2006; Lloyd and Hastings, 2009; Dysdale et al., 2009; Choi and Kovshoff, 2013; Woolfson et al., 2010; Armstrong and Dagnan, 2011). Two of the

studies did not provide specific details of the sample (Gale, 2009 and Gore and Umizawa, 2011). The other studies included small numbers of parents but also in some cases "family carers" (Gore and Umizawa, 2011) and grandparents (eg Whittingham et al., 2009). Finally, of those studies which included fathers, the sample size of fathers was always less than the sample of mothers and often represented less than 25% of the overall sample (eg, Whittingham et al., 2008).

Within the current studies, possibly due to the small numbers of fathers included, there have been no direct comparisons of findings between mothers and fathers. Hassall and Rose (2005) suggest that studies with parents of children with developmental disability show that similar levels of stress are experienced by both mothers and fathers. However, they also cite Dyson (1997) and state that different aspects of the family environment might influence the stress experienced by fathers compared to mothers and so a comparison of mothers and fathers would warrant further investigation. Failing to do so and analysing the data of mixed samples could be problematic. The inclusion of larger samples of mothers may reflect the tendency for caregiving roles to be fulfilled by mothers who tend to be the primary caregiver for their children with a developmental disability (DoH, 2008). As such, it seems that research based on mothers alone is useful in terms of our understanding of maternal perspectives but evidence also goes some way to supporting the notion that these findings can be generalised to fathers. Despite this, it would be a useful endeavour to fully explore the similarities and differences between mothers and fathers caring for a child with a developmental disability.

Population under examination: developmental disability

Another aspect of variability is the degree of developmental disability within each of the samples. A number of studies recruited parents of children with a developmental disability and do not provide information relating to the level of disability of the child (Lloyd and Hastings, 2009; Hassall et al., 2005; Hill and Rose, 2009; Armstrong and Dagnan, 2011), some include the parents of children with a severe to moderate developmental disability (Gore and Umizawa, 2011; Gale 2009; Drysdale et al., 2009), while a large proportion consider parents of children with Autism Spectrum Disorder (ASD; Meirsschaut et al., 2010; Hodgett wt al, 2013; Whittingham et al., 2009; Whittingham et al., 2006; Whittingham et al., 2008, Choi and Kovshoff, 2013; Dale et al., 2006) and one paper describes including parents of children with mixed developmental disabilities including specific genetic syndromes (Woolfson et al., 2010). However it is important to note that children and adults diagnosed with rare genetic syndromes comprise a significant proportion of people with intellectual disabilities (Oliver et al., 2010) and therefore all of the samples outlined above are likely to include children with either diagnosed or undiagnosed genetic syndromes. Given the wide variation of intellectual disability within the current review, it is difficult to generalise across findings. It could be hypothesised that the nature and presentation of different genetic syndromes, and/or level of disability of the child, may also impact upon the attributions parents make of their children's behaviour, and the current studies do not provide an exploration of this.

The emerging literature shows evidence of the development of models of parental cognitions however this is problematic when the samples under consideration are not

comparable. It is already generally accepted that level of disability, for example, influences outcome including parental mental health (Hassall and Rose, 2005; Emerson et al., 2004), and therefore it does not seem unreasonable to suggest that level of disability might also influence parental attributions. Caring for a child with Autism Spectrum Disorder (ASD) may be qualitatively different to caring for a child with severe or profound a developmental disability; this may influence parental emotional responses which may impact upon attributions. Evidence exists to support this proposition (Stratton and Swaffer, 1988, cited by Hassall and Rose, 2005) and Hassall and Rose (2005) summarise the research which has examined parental attributions in the parents of children with Attention Deficit Hyperactivity Disorder (ADHD). They suggest that these parents show distinctive patterns of attributions due to their beliefs that that ADHD has a biological basis. In the current review a number of the papers include children diagnosed with ASD; given that there is considerable uncertainty regarding the cause of ASD (Al Anbar et al., 2010) it could be hypothesised that the attributions made by parents in these studies may differ depending on the parents' beliefs about the cause of their child's ASD, such that those who believe that the ASD has a biological cause, may make different attributions to parents who have different beliefs about the causes. No methodological or statistical controls are made for this, resulting in possible bias in the findings.

Conceptualisation of Parenting Cognitive Variables

Appropriate Definition and measurement

The studies under review make reference to a range of concepts, which are defined and measured in different ways. In terms of parental cognitions, the current studies are similar to literature reviewed by Hassall and Rose in that the studies examine parental locus of control (Gale, 2009; Llloyd and Hastings, 2009), parental self efficacy (Hodgetts et al., 2013) and parental attributions (Armstrong and Dagnan, 2011; Choi and Kovshoff, 2013; Dale, Jahoda and Knott, 2006; Drysdale et al., 2009; Gore and Umizawa, 2011; Meirsschaut et al., 2010; Whittingham et al., 2006; Whittingham et al., 2008; Whittingham et al., 2009; Woolfson et al., 2010) and some of these studies have considered a combination of these (Lloyd and Hastings, 2009; Hassall et al., 2005; Hill and Rose, 2009). However, none has examined all three constructs simultaneously. As suggested by Hassall and Rose, these three concepts, whilst related, are conceptually unique with locus of control and parental self efficacy representing more general schema with parental attributions being considered more momentary and situation specific. As the literature is representing a broad focus of parental cognitions, and the fact that there has been no exploration in the current papers of the ways in which these three concepts relate to each other, this suggests absence of a clear conceptual framework.

Parental Attributions

Of the fifteen papers under review in the current study, all but two have assessed parental attributions of their child's behaviour. Of these thirteen papers, eight use Weiner's (1980) attributional model as a conceptual framework. Drysdale et al., (2009) used the framework as a method to develop a semi-structured interview for assessing maternal attributions of self injurious behaviour (SIB) and Dale et al., (2009) developed a semi-structured interview following a focus group whose themes had emerged consistent with the model. Of the remaining studies Weiner's (1980) dimensions of locus of cause, stability and controllability were measured using four different questionnaire measures. The *Attribution and Control questionnaire (ACQ)* was employed by Whittingham, Sofronoff and Sheffield, (2006) and was developed for the purposes of their study, the *modified attributional style questionnaire (ASQ;* Peterson et al., 1982 cited in Armstrong and Dagnan, 2011) was used by Armstrong and Dagnan (2011) and an adapted version of the *Parental Attribution Test (PAT*; Bugental, 2004 cited in Woolfson et al., 2010) was used by Woolfson et al., (2010). Three studies employed the *Parental Attribution Questionnaire* (*PAQ*; Whittingham et al., 2008) these were Whittingham et al (2008- the measure was developed for the purposes of this study), Whittingham et al., (2009) and Choi and Kovshoff (2013). The *PAQ* was also originally designed to incorporate Fishbein and Ajzen's (1975; as cited in Whittingham et al., 200) theory of reasoned action (that parents behaviour is strongly predicted by behavioural intentions). Each of the questionnaires is a vignette based questionnaire in which participants are presented with a scenario containing a child and a specific behaviour. The "child" and the "behaviour" in each scenario are manipulated depending on the research

question. Each of the measures is based on Weiner's (1980) attributional model, and some include Fishbein and Ajzen's (1975 cited in Whittingham et al., 2006) model of reasoned action (e.g.Whittingham et al., 2006). The *PAT* and the *PAQ* focus on attributions of parent and child internality, controllability and stability. The *ASQ* and the *ACQ* measure attributions of locus, stability and controllability. The face validity of all the measures described is good. However, none of these measures have been used alongside the other, so it is difficult to conclude whether they show robust convergent validity.

Within these studies a number of conclusions are drawn based on the *PAQ* in relation to child-referent attributions. However, these findings may be spurious given that the internal consistency of the child-referent locus was low for the good behaviour and ASD-related behaviour scenarios ($\alpha = 0.13$ and $\alpha = -0.06$ respectively). This issue is less concerning in the Choi and Kovshoff (2013) paper as they did not employ the good behaviour of ASD-related behaviour scales. They did however, adapt the questionnaire slightly to include prompts of types of disruptive behaviour.

To summarise, multiple measures of parental attributions based on Weiner's (1980) model are being employed. This results in a literature that lacks comparability and continues to be exploratory rather than confirmatory in nature. In order for the literature to begin moving forward, it would first seem necessary to develop a clear model which captures all aspects of parental cognition, at which time it would be helpful to develop and produce a robust measure of this which may, or may, not include parental attributions.

Parental Locus of Control and Self efficacy

Hassall and Rose (2005) noted that previous research tended to vary in terms of the way locus of control had been measured. In their review they found that often general measures of locus of control had been used and argued for the use of more specific parental locus of control measures. Within the current review, (Gale, 2009) used a structured interview that was designed to generate responses that might indicate a parent's locus of control. Three of the articles have used a specific locus of control measure, the *Parental Locus of Control Scale* (*PLOC*; Campis et al., 1986 cited by Lloyd and Hastings, 2009). These were Lloyd and Hastings (2009), Hassall et al., (2005) and Hill and Rose (2009) who all employed a shortened version of the scale. The *PLOC* was designed to measure five factors; parental self efficacy, responsibility, child's control of parent's life, parental belief in fate or chance and parental control of child's behaviour (Lloyd and Hastings, 2009). There does however, appear to be a degree of unreliability with this scale. In their study Lloyd and Hastings (2009) reduced the number of items in some of the subscales in order to achieve better Alpha levels, despite having already removed the items and scales that had been suggested by Campis et al., (1986, cited in Lloyd and Hastings, 2009). Furthermore, Lloyd and Hastings acknowledge that a number of variations of the *PLOC* have been used and that this presents difficulties and raises questions regarding the external and internal validity of this scale. As they suggest, it appears that the measure would benefit from further refinement in order to achieve a good and reliable measure of locus of control that can be used across samples.

Four of the studies within the current review specifically focus on efficacy (Meirsschaut et al., 2010; Hodgetts et al., 2013; Hassall and Rose, 2005; Hill and Rose, 2009). As recommended within the Hassall and Rose (2005) review each of these studies has employed a specific parental self-efficacy measure. Both Hassall et al., (2005) and Hill and Rose (2009) used the *Parenting Sense of Competence Scale (PSOC*; Gibaud-Wallaston & Wandersman 1978, as Hassall et al., 2005) whilst the *Parenting Self Efficacy Scale* was employed by Hodgetts et al., (2013) and finally Meirschautt et al., (2010) used the *Maternal Efficacy Scale* (Teti and Gelfand, 1991 cited by Meirschautt et al., 2010).

To summarise, since there has been a move forward in the measurement of parental self efficacy and locus of control, despite some of the difficulties outlined above. Future research should examine the links between parental self efficacy and locus of control. Presently it is unclear whether there are overlaps between these two constructs.

Parental Cognitive Factors- A Multi-Dimensional Model

Hassall and Rose recommended developing a multi-dimensional model of parental cognitive variables. This seems reasonable given that cognitive constructs have been shown to be linked to parental and child outcomes, including wellbeing of both, and also the likelihood of parents accessing support services. However, inspection of the current studies suggests that no single model is being developed; investigators are continuing to examine the individual constructs. The exception is Hill and Rose (2009), who examined both self-efficacy and locus of control in terms of the degree to which both constructs (termed “cognitive variables” in

the study) impact upon parental stress. They suggest their findings reveal that parental cognitive factors play an important role in the level of stress experienced by parents. Whilst this is a step forward in terms of developing understanding of the schematic aspects of parental cognitions, it seems that little attention has been paid to understanding how these are linked to individual momentary and situation specific attributions on parents of children with a developmental disability.

Defining Outcome variables

Within the current review outcome variables in each of the studies vary substantially. Some of the papers consider parental mental health; others define outcomes as changes in attribution. A number of studies include attribution as a predictor off parental stress/wellbeing (Hassall et al., 2005; Lloyds and Hastings, 2009; Meirschautt et al., 2010). Other studies have specifically examined the effectiveness or acceptability of training programmes using pre and post measures that include parental cognitive variables as well as parental wellbeing factors (Choi and Kovshoff, 2013; Gore and Umizawa, 2011; Hodgetts et al., 2013; Whittingham et al., 2009;). Woolfson et al., (2010) included parental controllability attributions as a moderator between developmental delay and problem behaviour.

Additionally, attribution is often defined and used in different ways within each of the studies. For example Woolfson et al., (2010) measured parental controllability attributions which describe one aspect of attribution based on Weiner's (1980) model, whereas other

studies have included all three dimensions from the original model (Whittingham et al., 2006). Again, this variability brings into question the generalisablity of each of the findings and how helpful they can be in developing a consistent multidimensional model which incorporates each of these parental cognitive factors. Thus varying outcomes within each of the studies limit the generalisability of the findings.

Parental Mental Health and Psychological Well-Being

Within the current review eight of the studies included measures (or discussions of measures in the case of qualitative methodologies) of psychological wellbeing or emotional responses of the parents (Armstrong and Dagnan, 2011; Dale et al., 2006; Drysdale et al., 2009; Gore and Umizawa, 2011; Hill and Rose, 2009; Hodgetts et al., 2013; Lloyd and Hastings, 2009; Meirsschaut et al., 2010). There is little consistency in terms of the measures used to capture aspects of parental emotional responses and/or wellbeing. Two studies attempt to measure parental emotional responses to children's challenging behaviour (Armstrong and Dagnan, 2001; Gore and Umizawa, 2011). Gore and Umizawa (2011) opted to use a measure previously developed that aimed to capture a parent's emotional response to a "gender neutral individual" displaying a broad range of challenging behaviours and this was measured prior to and post completing a training workshop and was used a measure of emotional change. The authors make no reference to the reliability of the original scale or the reliability within their study. Armstrong and Dagnan (2011) developed their own seven point likert scales, to capture feeling of anger and sympathy which they reported they based on emotions "highlighted by Weiner (1995)" (pp. 461, Armstrong and

Dagnan, 2011) and measured these in response to each of the behaviours under examination with good levels of internal consistency of these scales. Other studies also used a wide variety of measures such as the *Hospital Anxiety and Depression Scale* (*HADS*; Zigmond and Snaith, 1983), *Beck's Depression Inventory* (Beck, 1996 cited in Dale et al., 2006), *Nijmeegse Ouderlijke Stress Index* (De Brock et al., 1992 cited in Meirschautt et al., 2010), *Parenting Stress Index* (Abdin, 1995 cited in Dale et al., 2006), *Depression-Anxiety-Stress Scale* (Lovibond & Lovibond, 1995 cited in Hodgetts et al., 2013). Describing the nature of each of these scales is outside of the scope of the current review. However, this variability suggests that there is no consistency in terms of the way parental mental health is measured making it difficult to compare findings.

Whilst the current review did not include mental health/wellbeing or parental emotional responses as part of its search criteria, its inclusion in eight of the studies within this review may reflect acceptance that parental cognitive factors may be predictive of mental health. Furthermore, based on all of the factors outlined so far in the review, it is difficult to draw firm conclusions about the impact of parental attributions on parental mental health. However, within the current studies, patterns in terms of parental emotional responses and wellbeing do appear to mirror previous research into parental attributions and stress. For example, those parents who experienced a limited sense of control over their child's behaviour were likely to experience stress (Drsydale et al., 2009; Hill and Rose, 2009; Lloyd and Hastings, 2009) and also higher levels of parenting incompetence feelings were shown to be related to symptoms of depression (Meirsschaut et al., 2010). These would be

theoretically predictable relations based on a multi-dimensional model of parental cognitions.

Definitions of Behaviour

The third construct considered within the current review was children's behaviour. Only two of the studies took formal measures of children's problem behaviour using the *Child Behavior Checklist (CBCL*; Achenbach and Rescorla, 2001 cited and used by Woolfson et al., 2010) and the *Eyberg Child Behavior Inventory (ECBI*; Whittingham et al., 2009). Woolfson et al., (2010) report that within the original manual of the *CBCL* there was high internal reliability of the subscales and high test-retest reliability. Whittingham et al., (2009) used the *ECBI* a scale which was originally developed to measure the behaviour of typically developing children. However, they cite two studies that have utilised the scale with children diagnosed with Aspergers Syndrome. This means that it would not be possible to make comparisons between these two studies, but there is also a question of whether the *ECBI* used by Whittingham et al., (2009) is a valid measure for use with children with ASD.

Three studies (Hassall et al., 2005; Hill and Rose, 2009; Lloyd and Hastings, 2009) also employed a general measure of adaptive and non-adaptive behaviour (*VABS*: Sparrow et al., 2005, cited in Hill & Rose, 2009). In those studies where the VABS was employed, it was used to capture behavioural difficulties in the children and to assess the relationship with parenting stress (Hassall et al., 2005). These studies suggest that the presence of maladaptive behaviour in their children at least partly predicts parenting stress (Hill and

Rose, 2009). Given the low response rate within the Hill and Rose sample (20%), it could be argued that the findings in this particular study are not necessarily generalisable to parents of children with a developmental disability who do not present with behavioural difficulties.

Finally, the way that behaviour was considered in each of the papers varied a great deal. On a number of occasions the parents of children with "challenging", "problem" or "disruptive" behaviours were included within the study but no formal measure of this was taken (Dale et al., 2009; Gale, 2009) and another examined the parents of children with problem behaviours consistent with ASD, but again, took no formal measure (Choi and Kovshoff, 2013).

Conclusions

Parental Cognitions and their Relationship to Stress, Coping and Adaptation

Hassall and Rose (2005) made specific reference to models of stress and coping and stated that all are based on a multidimensional model; they suggest that in order to achieve this, parental cognitions need to be clearly conceptualised. However, despite this recognition, none of the studies within the current review consider parental coping in response to stress and child behaviours. Nor do any of the studies consider successful adaptation of parents to meet the needs of their children. The relationship between cognitions, coping and outcome, including mental health, is clearly established within the literature and it would be beneficial to include measures of coping in future research. There is a need for health professionals to understand the way parents are thinking about, experiencing and responding to the behaviours being presented by their children. As concluded by Hassall and Rose (2005) there continues to be, very little clear evidence about the way in which particular types of parental attributions impact upon outcome.

Furthermore, given that there is now an increasing acceptance that families often adapt to, and benefit from the experience of caring for a child with a developmental disability (Dura-Vila et al., 2010; Hassall and Rose, 2005; Hastings and Taunt, 2002; Horsley and Oliver, In press; Hyman and Oliver, 2001; Singer, 2006) Hassall and Rose suggested incorporating this into future investigations. However, within the current review, there was no use of quantitative measures of positive emotions, responses or experience. The Common Sense

Model of Illness Representations (Leventhal et al., 1980) would also incorporate positive outcomes, and provides further support for using this model in future research.

Defining Parental Cognition

"Parental cognitions" of locus of control, parental self efficacy and attributions are still being examined independently of each other. It seems that there is no model which incorporates locus of control, parental self efficacy and attributions, and other relevant cognitive factors. This raises the question of to what degree these concepts overlap and/or influence the other. Furthermore, when each of the concepts is under investigation there appears to be no consistent form of measurement, making comparisons between studies or populations difficult.

The current literature shows wide variation in terms of: aims, cognitive variables under measurement, population under examination, measurement tools employed and outcome variables. Given the impact that parents' cognitive responses can have on themselves and their children, it seems essential that a greater understanding of this is developed within the literature. As the majority of papers within the current review are either qualitative (and therefore exploratory in nature) or are quantitative within subjects designs, it is clear that the literature continues to be theory-model building rather than theory-model testing, suggesting that the examination of parental cognitions and of behaviour in their children with a developmental disability is still in its infancy.

Hassall and Rose (2005) summarise the research which has examined parental attributions in parents whose child's difficulties have a biological origin and suggest that these parents show distinctive patterns of attributions. This might suggest that in order to understand parental cognitions that relate to their child's difficulties, it may be helpful to gain an understanding of the parents beliefs about the origins of those difficulties. This also leads to the question, is the reason that we have no clear model or conceptualisation the result of the fact that parental cognition is not fully understood or defined, and so consequently the relationship between parental cognition and other variables cannot reliably be explored?

It also seems that there is a small amount of research aiming to explore and acknowledge the link between attributions and behaviour. The *PAQ*, developed by Whittingham (2006) was developed based in Weiner's (1980) model and also Fishbein and Ajzen's (1975 cited in Whittingham et al., 2006) theory of reasoned action; thus incorporating parental cognitive variables with behavioural responses. However, it seems that that there is no consensus regarding how these concepts can be clearly conceptualised, linked and understood within parents of children with a developmental disability.

In summary, there is no clear development of a model for understanding the complexity of parental cognitions and the ways that these impact upon parental mental health and wellbeing or any other potential outcome variable. These findings indicate that since Hassall and Rose (2005) suggested the development of such a model, which incorporates parental cognitive variables with coping and other outcomes, there have been limited advancements.

One way to move forward may be to draw upon models from the health literature which examine parental cognitions, behavioural responses and outcomes and emotional wellbeing. One example of this would be the common sense model of illness representations (Leventhal et al., 1980). This model addresses the way that cognitive factors influence illness coping behaviours and subsequent outcomes. It hypothesises that people develop a mental representation of an illness based on available information (media, friends and family and also personal experience of the illness) and this representation contributes to a person's understanding of the illness and guides the management of the illness threat. The "illness representation" comprises a cognitive and emotional representation. This model was originally developed and used in people experiencing poor health. Since its development, it has been adapted for use with parents and caregivers (Al Anbar et al, 2010, Barrowclough et al., Fortune et al, 1996) with initial findings suggesting that the model can successfully be applied to parents and caregivers and therefore provides an understanding of the way in which cognitive factors exert an influence over coping and wide ranging outcomes.

The Measurement of Parental Locus of Control

As recommended by Hassall and Rose (2005), there has been a move forward to using a parent specific locus of control measure. Within the current articles three of them utilised the *PLOC*. However, there remain questions around this particular measures reliability. This may be an issue with the reliability of the scale itself or reflect the variation in the samples within which it is used. Furthermore, there may be little use for this specific scale, until locus of control is understood in the context of a multidimensional model of parental cognitions, mental health and coping.

Clinical Implications

The clinical implications of not fully understanding parental cognitions, and the way that they relate to parental behaviours and child and parental wellbeing are potentially significant. Research has demonstrated that help-seeking behaviours can be predicted by parental cognitions (Hagger and Orbell, 2003). Furthermore, for children with developmental disability who are reliant upon caregivers to seek appropriate help and support at times of illness or difficulty, it is essential to understand the factors which may inhibit parents seeking support for their children, given the potential consequences of failing to do so. Finally, it would be beneficial to understand the ways in which parental cognitions influence coping responses and psychological wellbeing because in doing so, it will become possible for health professionals to develop appropriate interventions to support them.

References

Abdin, R.R. (1995) *Parenting Stress Index.* Psychological Assessment Resources: Florida, CA *cited in* Dale, E., Jahoda, A. & Knott, F. (2006). Mothers' attributions following their child's diagnosis of autism spectrum disorder. Exploring links with maternal levels of stress, depression and expectations about their child's future. *Autism, **10***, 463-479.

Achenbach, T. & Rescorla, L. (2001) Manual for the ASEBA School-Age Forms and Profiles. University of Vermont, Research Centre for Children, Youth and Families, Burlington, VT, USA. *Cited in* Woolfson, L.M., Taylor, R.J. & Mooney, L. (2011). Parental attributions of controllability as a moderator of the relationship between developmental disability and behaviour problems. *Child: Care, Health and Development, **32***, 184-194.

Al Anbar, N.N., Derdennes, R.M., Prado-Netto, A., Kaye, K. & Contejan, Y. (2010) Treatment choices in autism spectrum disorder: The role of parental illness perceptions. *Research in Developmental Disabilities, **31***, 817-828.

Armstrong, H. & Dagnan, D. (2011). Mothers of children with an intellectual disability: Their attributions, emotions and behavioural responses to their child's challenging behaviour. *Journal of Applied Research in Intellectual Disability, **24,*** 459-467.

Barker, C., Pistrang, N. And Elliott, R. (2002). *Research methods in clinical psychology: An introduction for students and practitioners (2nd Ed,).* Chichester: Wiley & Sons Ltd.

Beck, A., Steer, R.A., & Brown, G.K. (1996) *Manual for the Beck Depression Inventory*. Psychological Corporation: Texas *cited in* Dale, E., Jahoda, A. & Knott, F. (2006). Mothers' attributions following their child's diagnosis of autism spectrum disorder. Exploring links with maternal levels of stress, depression and expectations about their child's future. *Autism,* ***10:5***, 463-479.

Bugental, D.B. (2004) *Parent Attribution Test, Revised Manual.* Unpublished Measure. University of California: Santa Barbara. *Cited in* Woolfson, L.M., Taylor, R.J. & Mooney, L. (2011). Parental attributions of controllability as a moderator of the relationship between developmental disability and behaviour problems. *Child: Care, Health and Development,* ***32***, 184-194.

Bugental, D.B., Johnstone, C., New, M. & Silvester, J. (1998) Measuring parental attributions: conceptual and methodological issues. *Journal of Family Psychology,* ***12***, 459-480.

Campis, L.K., Lyman, R.D. & Prentice-Dunn, S. (1986) The parental locus of control scale: Development and validation*. Journal of Clinical Child Psychology,* ***15****,* 260-267*. Cited in* Lloyd, T. & Hastings, R.P. (2009) Parental locus of control and psychological wellbeing in mothers of children with intellectual disability. *Journal of Intellectual and Developmental Disability,* ***34***, 104-115.

Chavira, V., Lopez, S.R., Blacher, J., Shapiro, J. (2000) Latina mothers attributions, emotions, and reactions to the problem behaviours of their children with developmental disabilities, *Journal of Child Psychology and Psychiatry, **41**,* 245-252

Choi, K.Y.K. & Kovshoff, H. (2013) Do maternal attributions play a role in the acceptability of behavioral interventions for problem behaviour in children with autism spectrum disorders? *Research in Autism Spectrum Disorders, **7**,* 984-996.

Dale, E., Jahoda, A. & Knott, F. (2006). Mothers' attributions following their child's diagnosis of autism spectrum disorder. Exploring links with maternal levels of stress, depression and expectations about their child's future. *Autism, **10***, 463-479.

De Brock, A.J.L.L., Vermulst, A.A., Gerris, J.R.M., and Abidin, R.R. (1992). *Nilmeegse Ouderlijke Stress Index.* Swets en Zeitlinger BV: Lisse *cited in* Meirsschaut, M., Roeyers, H., & Warreyn, P. (2010) Parenting in families with a child with autism spectrum disorders and a typically developing child: mothers experiences and cognitions. *Research in Autism Spectrum Disorders, **4**,* 661-669.

Department of Health (2008). *Carers at the heart of the 21st century*. London: DoH

Drysdale, E.E., Jahoda, A. & Campbell, E. (2009) Investigating spontaneous attributions in mothers of individuals with intellectual disabilities and self-injurious behaviour. *British Journal of Learning Disabilities, **37***, 197-206.

Dyson, L.L. (1997) Fathers and mothers of school-age children with developmental disabilities: parental stress, family functioning and social support. *American Journal of Mental Retardation,* ***102*****,** 267-279.

Emerson, E., Roberston, J. & Wood, J. (2004) Levels of psychological distress experienced by family carers of children and adolescents with intellectual disabilities in an urban conurbation. *Journal of Applied Research in Intellectual Disability,* ***17***, 77-84.

Fishbein and Ajzen (1975) *Belief, attitude, intention and behaviour: an introduction to theory and research.* Philippines: Addison-Wesley Publishing Company Inc. Cited in Whittingham, K., Sofronoff, K. & Sheffield, J.K. (2006). Stepping Stones Triple P: A pilot study to evaluate acceptability of the program by parents of a child diagnosed with autism spectrum disorder. *Research in Developmental Disabilities,* ***27****,* 364-380.

Foster, R. H., Kozacehk, S., Stern, M. & Elsea, S.H. (2010) Caring for the caregivers: An investigation of factors related to well-being among parents caring for a child with smith-magenis syndrome. *Journal of Genetic Counselling,* ***19****,* 187-198.

Gale, I. (2009). "It's a tricky one to answer, that.." Locus of control in parents and caregivers of children with severe and complex learning disabilities and challenging behaviour. *Educational and Child Psychology,* ***26***, 31-42.

Gore, N. & Umizawa, H. (2011) Challenging behaviour training for teaching staff and family carers of children with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, **8**,* 266-275.

Gibaud-Wallston, J. & Wandersman, L.P. (1978) *Development and Utility of the Parenting Sense of Competence Scale.* Paper presented at the meeting of the American Psychological Association, Toronto. *Cited by* Hassall, R., Rose, J. and MacDonald, J. (2005). Parenting stress in mothers of children with an intellectual disability: the effects of parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research, **49,** 405-418*

Hagger, M.S. & Orbell, S. (2003) A meta-analytic review of the common-sense model of illness representations. *Psychology and Health, **18***, 141-184.

Hassall, R. & Rose, J. (2005). Parental cognitions and adaptation to the demands of caring for a child with an intellectual disability: A review of the literature and implications for clinical interventions. *Behavioural and Cognitive Psychotherapy, **33***, 71-88.

Hassall, R., Rose, J. and MacDonald, J. (2005). Parenting stress in mothers of children with an intellectual disability: the effects of parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research, **49,** 405-418*

Hastings, R. & Taunt, H. (2002) Positive perceptions in families of children with intellectual disabilities. *American Journal of Mental Retardation, **107***, 116-127

Hill, C. & Rose, J. (2009) Parenting stress in mothers of adults with an intellectual disability: parental cognitions in relation to child characteristics and family support *Journal of Intellectual Disability Research, **53***, 969-980.

Hodgetts, S., Savage, A. & McConnell, D. (2013) Experience and outcomes of stepping stones triple P for families of children with autism. *Research in Developmental Disabilities, **34***, 2572-2585.

Horsley, S. and Oliver, C. (in press) Positive impact and its relationship to wellbeing in parents of children with intellectual disability: a literature review. Submitted to *British Journal of Developmental Disorders.*

Johnson, R.F., O-Reilly, M. & Vostansis, P. (2006) Caring for children with learning disabilities who present problem behaviours: a maternal perspective. *Journal of Child Health Care, **10*****,*** 188-198.

Johnstone, C., Hessl, D., Blasey, C., Eliez., S., Erba, H., Dyer-Friedman, J., Glaser, B. & Reiss, A.L. (2010). Factors associated with parenting stress in mothers of children with fragile X syndrome. *Developmental and Behavioral Paediatrics, **24*****,*** 267-275.

Kim, H.W., Greenberg, J.S., Seltzer, M.M., & Krauss, M.W. (2003). The role of coping in maintaining the psychological wellbeing of mothers of adults with intellectual disability and mental illness. *Journal of Intellectual Disability Research,* ***47,*** 313-327.

Lazarus and Folkman (1984) *Stress, appraisal and coping*. New York: Springer *cited by* Hassall, R. & Rose, J. (2005). Parental cognitions and adaptation to the demands of caring for a child with an intellectual disability: A review of the literature and implications for clinical interventions. *Behavioural and Cognitive Psychotherapy,* ***33***, 71-88.

Leventhal, H., Meyer, D & Nerenz, D. (1980). The common sense model of illness danger. In S. Rachman (Ed.) *Medical Psychology*, Vol 2. pp 7-30. Pergamon, New York.

Leventhal, H., Benyamini, Y., Brownlee, S., Diefenbach, M., Leventhal, E., Patrick-Miller, L. & Robitaille, C. (1997). Illness representations: Theoretical foundations. In: K. J. Petrie, & J. Weinman (Eds.), *Perceptions of Health and Illness: Current Research and Applications* (pp. 19–45). Amsterdam: Harwood Academic Publishers.

Lloyd, T. & Hastings, R.P. (2009) Parental locus of control and psychological wellbeing in mothers of children with intellectual disability. *Journal of Intellectual and Developmental Disability,* ***34***, 104-115.

Lovibond, S.H. & Lovibond, P.F. (2005). *Manual for the Depression Anxiety Stress Scales* (2nd Ed.) Psychology foundation: Sydney *cited in* Hodgetts, S., Savage, A. & McConnell, D. (2013) Experience and outcomes of stepping stones triple P for families of children with autism. *Research in Developmental Disabilities,* ***34***, 2572-2585.

Mash, E.J. and Johnstone, C. (1990) Determinants of parenting stress : illustrations from families of hyperactive children and physically abused children. *Journal of Clinical Child Psychology,* ***19*****,** 313-328.

McCubbin, H. I., & Patterson, J. M. (1983). The family stress process: the double ABCX model of adjustment and adaptation. In H.I. McCubbin, M.B. Sussman and J.M. Patterson (Eds), *Social Stress and the Family: Advances and Developments in Family Stresses Theory and Research* (pp. 7-37) *cited in* Hassall, R. & Rose, J. (2005). Parental cognitions and adaptation to the demands of caring for a child with an intellectual disability: A review of the literature and implications for clinical interventions. *Behavioural and Cognitive Psychotherapy,* ***33***, 71-88.

Meirsschaut, M., Roeyers, H., & Warreyn, P. (2010) Parenting in families with a child with autism spectrum disorders and a typically developing child: mothers experiences and cognitions. *Research in Autism Spectrum Disorders,* ***4*****,** 661-669.

Morrisey-Kane, E. & Prinz, R.J. (1999). Engagement in child and adolescent treatment: the role of parental cognitions and attributions. *Clinical Child and Family Psychology Review, **2***, 183-198)

Oliver, C., Woodcock, K. & Adams, D. (2010). *The importance of aetiology of intellectual disability.* In: Grant, G., Ramcharan, P., Flynn, M. and Richardson, M. (Eds.) Learning Disability: A life cycle approach to valuing people. (pp. 135-146). Open University Press: Wiley.

Padeliadu, S. (1998) Time demands and experienced stress in Greek mothers of children with down's syndrome. *Journal of Intellectual Disability Research, **42***, 144-153.

Peterson, C., Semmel, A., von Baeyer, C., Abramson, L.Y., Metalsky, G.I. & Seligman, M.E.P. (1982) The attributional style questionnaire. *Cognitive Therapy and Research**, 6,*** 287-299*. Cited in* Armstrong, H. & Dagnan, D. (2011). Mothers of children with an intellectual disability: Their attributions, emotions and behavioural responses to their child's challenging behaviour. *Journal of Applied Research in Intellectual Disability, **24,*** 459-467.

Rotter, J.B. (1966). Generalized expectancies for internal versus external control of reinforcement. *Psychological Monographs*, 80:1, 1-28.

Sparrow, S.S., Ciccetti, D.V. & Balla, D.A. (2005). *The Vineland Adaptive Behaviour Scale (Interview edition survey form)*. American Guidance Service: MN *cited in* Hill, C. & Rose, J. (2009) Parenting stress in mothers of adults with an intellectual disability: parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research, **53***, 969-980.

Stratton, P. & Swaffer, R. (1988) Maternal causal beliefs for abused and handicapped children. *Journal of Reproductive and Infant Psychology, **6,*** 201-216. *Cited in* Hassall, R. & Rose, J. (2005). Parental cognitions and adaptation to the demands of caring for a child with an intellectual disability: A review of the literature and implications for clinical interventions. *Behavioural and Cognitive Psychotherapy, **33***, 71-88.

Teti, D.M. & Gelfand, D.M. (1991). Behavioral competence among mothers of infants in the first year: the meditational role of maternal self efficacy. *Child Development, **62,*** 918-929. *Cited in* Meirsschaut, M., Roeyers, H., & Warreyn, P. (2010) Parenting in families with a child with autism spectrum disorders and a typically developing child: mothers experiences and cognitions. *Research in Autism Spectrum Disorders, **4*,** 661-669.

Wulffaert, J., van Berckelaer-Onnes, I., Kroonenberg, P., Scholte, E. & Hennekam, R. (2009) Simultaneous analysis of the behavioural phenotype, physical factors and parenting stress in people with cornelia de lange syndrome. *Journal of Intellectual Disability Research, **53***, 604-619.

Weiner, B. (1980) A cognitive (attribution)-emotion-action- model of motivated behaviour: an analysis of judgements of help-giving. *Journal of Personality and Social Psychology, **39***, 186-200.

Weiner, B. (1985) An attributional theory of achievement motivation and emotion. *Psychology Review, **92**,* 548-573.

Whittingham, K., Sofronoff, K. & Sheffield, J.K. (2006). Stepping Stones Triple P: A pilot study to evaluate acceptability of the program by parents of a child diagnosed with autism spectrum disorder. *Research in Developmental Disabilities, **27**,* 364-380.

Whittingham, K., Sofronoff, K., Sheffield, J.K. & Sanders, M.R. (2008) An exploration of parental attributions within the autism spectrum disorders population. *Behaviour Change, **25**,* 201-214.

Whittingham, K., Sofronoff, K., Sheffield, J.K. & Sanders, M.R. (2009) Do parental attributions affect treatment outcome in a parenting programme? An exploration of the effects of parental attributions in an RCT of stepping stones triple P for the ASD population. *Research in Autism Spectrum Disorders, **3**,* 129-144.

Woolfson, L.M., Taylor, R.J. & Mooney, L. (2011). Parental attributions of controllability as a moderator of the relationship between developmental disability and behaviour problems. *Child: Care, Health and Development, **32***, 184-194.

Zigmond, A.S. and Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica, 67*, 361-370.

Chapter 2- Empirical Paper

Parental Perceptions of Children's Genetic Syndromes and their Impact on Coping and Mental Health

Abstract

Background. Parental perceptions of their child's genetic syndrome were explored using the Common Sense Model of Illness Representations (Leventhal et al., 1980). Associations between parental perceptions, coping behaviours, affect and mental health were explored, including assessing whether level of disability and age of child impacted upon perceptions.

Method. One hundred and thirteen participants completed the Illness Perceptions Questionnaire Revised for Genetic Syndromes (IPQ-RGS). They also completed measures of positive and negative affect, anxiety and depression and coping behaviours.

Results. Inter-correlations between the IPQ-RGS subscales support the Common Sense Model of Illness Representations. Psychological outcomes for parents can in part be predicted by illness perceptions and coping strategies; a number of significant relationships were observed between illness representations, coping and psychological outcomes. Regression analyses revealed predictable relations based on the Common Sense Model, between illness perceptions, coping and psychological wellbeing.

Conclusions. Findings provide preliminary support for the hypothesis that the Common Sense Model of Illness Representations can be applied to the caregivers of children with rare genetic syndromes. Significant relationships were observed between illness representations, coping and psychological wellbeing suggesting that illness representations may have important implications for coping strategies and caregiver wellbeing.

Keywords: Rare genetic syndromes, parents, perceptions, coping, affect, anxiety and depression

Introduction

Raising a child and caring for an adult with an intellectual disability is associated with some negative psychological consequences for many carers (DoH, 2008). A number of child and adults variables are associated with these consequences, including the level of intellectual disability (Emerson et al., 2004), associated health problems (Berg et al., 2007) and presence of challenging behaviour (Hassall, Rose & McDonald, 2005; Herring, 2006; Hastings et al., 2006; Orsmund et al., 2002). Given the importance of these variables, exploration of other characteristics might reveal additional influences that should also be further investigated. Children and adults diagnosed with rare genetic syndromes comprise a significant proportion of people with intellectual disabilities (Oliver and Woodcock, 2008) and it is possible that the cause of intellectual disability could exert an influence on the relationship between child characteristics and parental wellbeing.

Research into the impact of raising a child with a rare genetic syndrome on parental wellbeing, is in its infancy. However, given that people with rare genetic syndromes make up a significant proportion of people with intellectual disability (Oliver and Woodcock, 2008), it is reasonable to suggest that the literature which has focused on the parents of children with intellectual disability may also reflect the experiences of parents of children with rare genetic syndromes. Parents of children with an intellectual disability are faced with many social and practical problems that can lead to significant parental distress (Dura-Vila et al., 2010; Hill & Rose, 2009; Kim et al., 2003; van der Borne et al., 1999). It is therefore unsurprising that caregivers of children diagnosed with a genetic syndrome often experience

elevated levels of common mental health problems such as anxiety and depression (Griffith et al., 2011; Horsley and Oliver, In press; Norizan and Shamsuddin, 2010).

Psychological Outcomes for Carers

It has long been recognised that caregivers of children with intellectual disability are at increased risk of experiencing depression compared to the parents of typically developing (TD) children (Singer, 2006). Research has also focused on stress and anxiety in caregivers of children with intellectual disability (Miller et al., 1992) and this has more recently been extended to investigate parents of children with genetic syndromes. These findings suggest stress and/or anxiety levels is also elevated in these caregivers (Foster et al., 2010; Johnstone et al., 2010; Padeliadu, 1998; Wulffaert et al., 2009).

However, some comparison studies have revealed no significant differences in levels of depression in parents of children with intellectual disability compared to parents of typically developing children and this, Singer (2006) suggests, reflects the emerging view that parental responses to rearing a child with an intellectual disability are complex and include positive outcomes and adaptive responses (Dura-Vila et al., 2010; Hassall and Rose, 2005; Hastings and Taunt, 2002 Horsley and Oliver, In press; Hyman and Oliver, 2001; Singer, 2006). In their review of the literature, Horsley and Oliver (In press) suggest that the positive impact of caring for a child with intellectual disability on parental psychological wellbeing, may exert a mediating effect between gain in the care giving experience and the negative affect associated with stress and depression. They state that positive impact refers to emotions, perceptions, reflections or relationship dynamics that are welcomed,

advantageous or constructive which occur directly as a result of caring for a child with intellectual disability. They concluded that the positive impact of caring for a child with intellectual disability may "buffer" the effect of stress and enhance parental wellbeing.

Psychological Wellbeing of Parents of Children with Rare Genetic Syndromes

There have now been a number of studies which have focused on the psychological wellbeing of parents of children with rare genetic syndromes. Johnston et al., (2010) found that mothers of children with fragile X Syndrome (fraX) had higher levels of parenting stress compared to the normative population. They also found that child characteristics, such as behavioural problems, were the main contributors to stress. Similar findings were also reported by Foster et al., (2010) who found that in caregivers of children with Smith-Magenis Syndrome (SMS) there were elevated levels of both anxiety and depression. Wulffaert et al., (2009) investigated "parenting stress" in the parents of children with Cornelia de Lange Syndrome (CdLS) and hypothesised that parenting stress might be related to the presence of self-injurious behaviour (SIB). However, their findings revealed that parenting stress was higher when the person with CdLS presented with behavioural problems but not SIB specifically or the severity of physical characteristics alone. Norizan and Shamsuddin (2010) also found parenting stress to be related to behavioural problems in children with Down syndrome. To summarise, these findings suggest that caring for a child with a genetic syndrome can result in caregivers experiencing elevated levels of stress and anxiety and that the experience of this might be influenced by factors such as behavioural problems in their children.

Parental Cognitive Factors

Given that previous research has shown elevated levels of anxiety and depression in the carers of children with rare genetic syndromes, it is important to consider the cognitive factors which may impact upon the development of these difficulties. Hassall and Rose (2005) suggest that there is general agreement in the literature about the importance of parental cognitions in relation to their impact upon parental wellbeing. Parental cognition in this context refers to appraisals, the meaning that is assigned to important events, along with parental beliefs about themselves and their children.

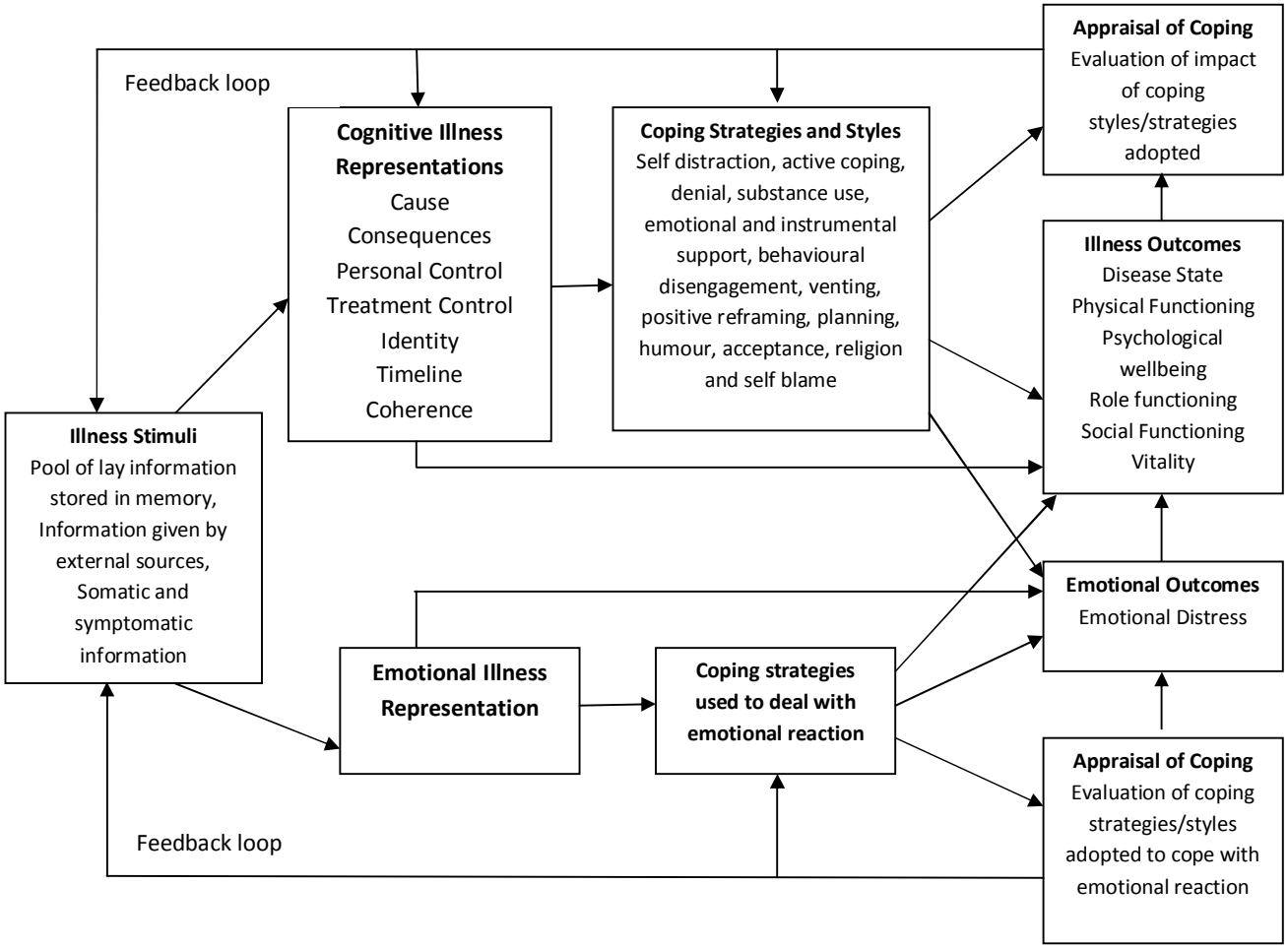
The Common Sense Model of Illness Representations

In addition to having a role in psychological wellbeing it is also important to consider the way in which parental cognitive factors may impact upon help seeking behaviours for the difficulties associated with their child's genetic syndrome. Within the physical health domain, research has focused on understanding the factors which influence adherence to medical regimes and on health behaviours related to the management of illness (Hagger and Orbell, 2003). Based on the findings within the physical health literature, which has found that illness representations guide helping seeking and coping responses (Hagger and Orbell, 2003), it could be hypothesised that caregiver's representation of their child's genetic syndrome may guide help seeking behaviours and subsequently impact upon their own emotional wellbeing. Difference is observed between individuals in the way they think about and respond to health conditions and it is by understanding these differences that targets for intervention may be identified (Hagger and Orbell, 2003). A number of models and theories have been developed to aid understanding of this process including the

common sense model of illness representations (also commonly referred to as the self-regulatory model; Leventhal et al., 1980).

The common sense model of illness representations addresses the way that cognitive factors influence illness coping behaviours and subsequent outcomes. It hypothesises that people develop a mental representation of an illness based on available information (media, friends and family and also personal experience of the illness) and this representation contributes to a person's understanding of the illness and guides the management of the illness threat. The "illness representation" comprises a cognitive and emotional representation. Hagger and Orbell (2003) highlight this dual processing, such that people will make simultaneous cognitive and emotional representations of an illness and both of these aspects will determine outcome. These representations guide the approach to coping which in turn impact upon illness and emotional outcomes. The model then hypothesises that the coping strategies adopted will be evaluated by the person in terms of their effectiveness in helping coping, which will further impact upon both emotional and illness outcomes. Hagger and Orbell (2003) provide a schematic representation of these relationships (See Figure 1).

Figure 1: Hagger and Orbell (2003)'s Schematic representation of Leventhal et al.'s (1980) Common Sense Model of Illness representations.



Illness representations can be organised into the dimensions of: cause, consequence, identity and timeline (Hagger and Orbell, 2003; Meyer et al., 1985). "Cause" refers to the beliefs that a person holds about what has been responsible for causing the illness, "Consequence" to the beliefs about the degree to which the illness will impact upon quality of life and ability to function effectively (Hagger and Orbell, 2003; Moss-Morris et., 2002), and "Identity" to the beliefs about the label attached to the illness and also their knowledge about the symptoms relating to the illness. Finally, "timeline" refers to beliefs about the likely duration of the illness and its symptoms.

A great deal of research has focused upon the applicability of the common sense model of illness representations to physical illness or disease. Within their meta-analytic review, Hagger and Orbell (2003) identified studies which examined the applicability of the common sense model to 23 different illnesses including chronic fatigue syndrome, irritable bowel syndrome, Diabetes type 1 and 2, psoriasis, muscular-skeletal injuries, cervical abnormalities and coeliac disease (Hagger and Orbell, 2003). Each of the studies provided empirical support for the application of the common sense model. Findings from the meta-analysis suggest "theoretically predictable relationships between illness cognitions, coping and outcome" (pg 176, Hagger and Orbell, 2003). These relationships were consistent with hypotheses based on Leventhal et al.'s (1980) common sense model. For example, common sense model dimensions will be linked directly to the strategies that a person adopts to cope with the illness threat (Leventhal et al., 1980). Hagger and Orbell (2003) state that the model implies a causal relationship such that the illness representation will influence the coping behaviour selected and that this will be in proportion to the perceived severity of the

illness threat. Studies are now emerging which have examined this in relation to parental/caregiver perceptions of their child's illness and have applied the common sense model to parental carers of people with schizophrenia (Barrowclough et al., 2001; Fortune et al., 2005) and autism spectrum disorders (Al Anbar et al., 2010). The findings of these three studies reflect findings in the physical health literature which provide support for the applicability of the common sense model more widely.

Capturing Illness Representations- The Illness Perceptions Questionnaire.

The Illness Perceptions Questionnaire (IPQ: Weinman et al., 1996) was developed originally to capture the constructs identified within the common sense model of illness representations. The scale was developed further to include additional subscales which resulted in the *Illness Perceptions Questionnaire-Revised (IPQ-R*) (Moss-Morris et al., 2002). A number of studies have adapted the scale for use with caregivers, family and paid staff (Al Anbar et al. 2010; Barrowclough et al., 2001; Fortune et al., 2005; Williams and Rose, 2007). Modifications have largely been made to the identity subscale, as the symptoms must have relevance to the illness/condition. Also, the *IPQ-R* was designed to assess a person's perception of their own illness, so items were reworded to refer to the participant's perceptions of the illness experienced by the person for whom they care. Initial evidence suggests that adapting the scale for use with caregivers has been successful (Fortune et al., 2005; Williams and Rose 2007) and provides support for the application of the model to caregivers of children with rare genetic syndromes.

To summarise, research has shown that genetic syndromes are associated with significant physical health difficulties (Berg et al., 2007), intellectual disability (Oliver et al., 2010) and various forms of challenging behaviour (Arron et al., 2011; Bull et al., 2011; Hayes et al., 2011; Sloneem et al., 2011; Teixeira et al., 2011). It has been demonstrated that these difficulties are associated with poor outcomes for parental carers of children with rare genetic syndromes (Foster et al., 2010; Johnstone et al., 2003; Norizan and Shamsuddin, 2010; Wulffaert et al., 2009). Illness representations are likely to impact upon coping response which will exact a mediating effect on psychological outcome (Hagger and Orbell, 2003). However, to date research has yet to apply the common sense model of illness representations to parents of children with genetic syndromes associated with intellectual disability.

Aims and Hypotheses

The current study will examine caregiver illness representations of their child's genetic syndrome in order to describe the theoretically predictable relations demonstrated in previous literature and explore the impact of the illness representation on caregiver coping, mental health and wellbeing. More specifically, the study will examine if Leventhal's (1980) common sense model of illness representations can be applied to the parents of children with rare genetic syndromes to predict parental psychological distress. Parental illness representations will be captured with a modified version of the *IPQ-R; the Illness Perceptions Questionnaire- Revised for Genetic Syndromes (IPQ-RGS*). Internal consistency of modified

scales will be assessed. It is predicted that relationships within the *IPQ-RGS* will evidence a pattern of associations between illness perceptions, coping styles and psychological wellbeing outcomes that is similar to those reported in previous studies.

The study aims to describe the degree of endorsement of child characteristics made by parents in the current sample (as measured by the IPQ-RGS identity subscale) and provide a descriptive summary of the average scores on each of the measures by the current sample. Two hypotheses are proposed: 1) caregivers levels of anxiety, depression and positive and negative affect will be related to illness representations and coping styles and 2) illness representations will be associated with the coping behaviours endorsed. The latter will be consistent with previous research findings, such that: a) control/cure illness representations will be positively related to problem-focused coping strategies (for example planning and active coping) and b) illness representations of consequence, identity and timeline will be positively related to the expression of emotions and/or avoidance or denial coping strategies (Hagger and Orbell, 2003).

Method

Participants

The sample consisted of a subgroup of parents/carers recruited to a larger study investigating the behavioural functioning of children and adults diagnosed with rare genetic syndromes (e.g. Arron et al., 2011; Burbidge et al., 2010; Moss et al., 2009). 387 parents and carers of people with a rare genetic syndrome, who had given written consent to be contacted about future research, were contacted by letter or e-mail (see Appendix 2). Participants were asked to take part in an online survey and were provided with written information about the study and a telephone number and e-mail address for further information (see Appendix 1). All participants had provided written informed consent. A total of 113 (29.20%) participants agreed to take part. Demographics presented within the results section are based on 106 participants. The demographic data for seven participants were not available due to their not taking part in previous studies.

Participants were the parents of children with a diagnosed genetic syndrome aged under 16. The following descriptive information is based on 78% (n= 88) of the total sample as data for the remaining 22% were unavailable. Participants were aged between 23 and 59 years of age (mean = 41.62, SD = 6.59) and 86% (76) were mothers, 9% (8) were fathers and the remaining 5% consisted of one adoptive mother, one grandmother and two foster carers. Of the sample, 98% (86) had their child living at home with them and the age of the child of the participants ranged from 2 to 15.11 years of age (mean = 9.96, SD = 4.09). Participants' children had been diagnosed with the following genetic syndromes: Angelman Syndrome

(n= 32), Cri Du Chat (13), CdLS (9), Prader-Willi Syndrome (15), Lowe Syndrome (3), Smith Magenis Syndrome (10), Sotos (6), 1p36 deletion (13), 9q34 deletion (2), 8p23 deletion (5) and Phelan McDermid Syndrome (5). Table 3 provides descriptive information of the child's level of disability based on the *Wessex* scores (Kushlick et al., 1973).

Table 3. Frequency and percentage of *Wessex* categorisation of children's level of disability

	N	**Category**	**Frequency**	**Percentage**
Hearing	93	*Deaf/Poor*	*22*	*23.7*
		Normal	71	76.3
Vision	89	*Blind/poor*	*31*	*38.4*
		Normal	58	65.2
Speech	94	*Non-Verbal*	*30*	*31.9*
		Verbal/Partly Verbal	64	68.1
Self Help Category	94	*Not Able*	*48*	*51.1*
		Partly Able/Able	46	48.9
Mobility Category	92	*Non/Partly Ambulant*	*38*	*41.3*
		Ambulant	53	58.7

Measures

Participants completed the following questionnaires:

Illness Perception Questionnaire- Revised for Genetic Syndromes (IPQ-RGS)

The *IPQ-RGS* is a modified version of the *Illness perceptions questionnaire-revised (IPQ-R*: Weinman et al., 1996- see Appendix 3). The *IPQ-R*, in its original form, consists of seven scales used to assess illness components that have been derived from the physical health literature and is based on Leventhal's common sense model of illness representations (Leventhal et al., 1980; Leventhal et al., 1997). These components are: cause, identity, timeline acute/chronic, timeline cyclical, consequences, personal control, treatment control, illness coherence and emotional representations. The causal scale was omitted from the current study as the cause is self-evident. The identity scale describes the caregiver's ideas about the label attached to their child's genetic syndrome and how this relates to the caregivers perception of the child's symptom experience. The cause scale refers to a person's perception of what has caused their illness, whilst both time-line scales refer to the person's perception of the likely duration of the illness and its symptoms; this has been categorised further in scales of acute/chronic and cyclical. The consequences scale refers to the person's perception of severity of the illness and the impact on areas of their functioning including physical, social and psychological. Finally, two control scales of treatment and personal control which describes the person's perceptions of the degree to which the illness can be controlled and/or cured by treatments or by actions of the individual. The illness coherence scale is concerned with how the illness "makes sense as a whole to the patient" (Weinman et al., 1996). For the purpose of the identity scale, participants indicate if they

have experienced twelve symptoms and they are then asked to indicate if they believe that symptom is related to their illness. All other scales ask participants to indicate their agreement with a statement on a five point scale from "strongly agree" to "strongly disagree. The *IPQ-R* was designed to assess a patient's perception relating to their own physical illness. As a result, there were a number of adaptations made for the purposes of the current study. Firstly, items were reworded so that the items referred to the parent's perceptions of how their child's genetic syndrome impacted upon their child and so that the personal control scale related to the degree to which the participant felt that they were able to control the symptoms/characteristics of their child's genetic syndrome. Secondly, the timeline acute chronic was removed as it was felt not relevant for the current population given that parents of children with a genetic syndrome are aware that the condition is lifelong.

Validity and Internal Reliability of the IPQ-RGS Identity Subscale

In line with the development of the original *IPQ-R* (Moss-Morris et al., 2001), the validity of the identity subscale was tested in two ways, firstly using a paired sample's t-test to examine differences within the scale between identity and somatisation and secondly via inspection of the frequencies of endorsements to assess face validity of the list of characteristics/symptoms included in the scale (Moss-Morris et al, 2001). A paired samples *t*-test revealed a significant difference between the number of characteristics identified as being experienced by the participants' child and the number of those characteristics that were attributed to genetic syndrome of the child (t (2.42), $p < 0.01$; see Appendix 4). This test therefore demonstrates that the identity scale developed for the purposes of the

current study captures identity as opposed to somatisation and demonstrates acceptable internal consistency (Cronbach's α of .65).

Internal Consistency of the IPQ-RGS Subscales

Cronbach's alpha was used to assess the internal consistency of each of the *IPQ–RGS* scales. Cronbach's alpha is one of the most widely used measures of internal consistency and assesses the degree to which each of the items within a given scale are measuring a single construct (Williams and Rose, 2007). In order for internal consistency to be considered acceptable, values should be 0.7-1 (with scores ranging from 0-1) (DeVellis, 1991, cited by Al Anbar, 2010). However, some authors have suggested that scores of 0.6 upwards can be considered acceptable (Al Anbar et al, 2010).

Initial inspection of the *IPQ-RGS* subscales revealed α values that suggested robust internal consistency of all scales with the exception of personal control. In order to improve the internal consistency of this scale, one item (number 12) was removed to increase the Cronbach α values to above the desired 0.7 cut off. The scales identity (α = 0.65), consequences (α = 0.67), and treatment control (α = 0.67) can be considered acceptably robust whilst the remaining scales show very good internal consistency (personal control: α = 0.78, illness coherence: α = 0.82, timeline cyclical: α = 0.82 and emotional representation: α = 0.85).

Brief COPE (Carver, 1997).

The B*rief COPE* is a 28 item shortened version of the original *COPE* scale developed by Carver et al., (1989; see Appendix 7). The *brief COPE* removed two scales of the original version and reduced each of the scales to two questions (Carver, 1997). The *brief COPE* therefore consists of 14 subscales, each containing two items. Participants rate the extent to which they use a coping strategy by indicating either "I haven't been doing this at all" (score of 1), "I've been doing this a little bit" (score of 2), "I've been doing this a medium amount" (score of 3) and "I've been doing this a lot" (score of 4). At the point of development Carver stated that all scales of the *Brief COPE* were at least minimally acceptable in terms of internal reliability with three of the scales having an α value of 0.50-0.59, with all remaining scales have α values of 0.60 upwards (Carver, 1997).

The Hospital Anxiety and Depression Scale (*HADS*; Zigmond and Snaith, 1973).

The *Hospital Anxiety and Depression Scale*, developed by Zigmond and Snaith (1973; see Appendix 8) was used to examine self-report levels of anxiety and depression. The items are rated on a scale of 0 to 3 and indicate the degree to which the participant agrees with a given item. This gives a maximum total of 21 for each of the subscales. Zigmond and Snaith also provide details of clinical cut off points in terms of severity of presentation based upon the composite scores for both.

The Positive and Negative Affect Scale (PANAS; Watson et al., 1988).

The *PANAS (*see Appendix 9) was originally developed to measure positive and negative affective state. Each scale consists of 10, one word items. Participants rate each item on a

five point scale where 1 = very slightly or not at all and 5 = extremely for their mood over the previous fortnight. This time frame was selected to be comparable to the time frame used within the *HADS*. The *PANAS* has been shown to have excellent internal reliability (Watson et al., 1988).

The Wessex (Kushlick et al., 1973).

The Wessex (see Appendix 10) measures both social and physical capacity of people with disabilities. Carers answer a number of questions which relate to physical capacity including vision, hearing and mobility, and social capacity such as communication, literacy and self care. Since its development the validity and reliability of scale have been robustly supported (Oliver et al., 2008). Higher scores on this measure are an indication of greater level of ability.

Data Analysis

The current study aimed to assess whether Leventhal et al.'s (1980) common sense model of illness representations can be applied to the caregivers of children with a rare genetic syndrome. In order to do this regression analyses were conducted. To determine the appropriate method of regression, the data were initially assessed for normality then univariate associations with anxiety and depression, positive and negative affect were assessed and four logistic regression analyses were performed with selected *IPQ-RGS* and *Brief COPE* scales as independent variables onto anxiety, depression, positive and negative affect. Due to the high number of independent variables the significance criterion was set at $p<0.01$ in order to limit type 1 errors.

Tests for Normality

The Kolmogorov-Smirnov test for normality of each of the scales along with inspection of the histograms suggested that the variables were not normally distributed and logistic regression was selected for analysis of the data. To perform this analysis it was first necessary to split the sample into groups. Due to the slight differences in univariate associations with anxiety, depression, positive and negative affect each was entered into separate regression analyses.

Results

Psychological Wellbeing in Carers

In order to test the first hypothesis, and given that the data were to be subjected to logistic regression, it was first necessary to split the sample into groups. For anxiety and depression the sample was split into 'normal' and clinical levels. Table 4 shows the frequency of participants in each of the *HADS* categories for anxiety and depression and how groups were formed for the purpose of the analysis. Using the cut-off to identify clinical presence of anxiety and depression according to the *Hospital Anxiety and Depression Scale* (*HADS; Zigmond and Snaith, 1983*) 66% of carers (n = 75) were experiencing anxiety and 35% (n = 39) were experiencing depression.

Table 4. Frequency of *HADS* Anxiety and Depression Scores by category

	N	Category	N	Percentage	Category	N	Percentage
Anxiety	113	Normal	38	33.6	Normal	38	33.6
		Mild	*40*	*35.4*	*Clinical levels*	*75*	*66.4*
		Moderate	*16*	*14.2*	*(Mild/Moderate*		
		Severe	*19*	*16.8*	*and Severe)*		
Depression	113	Normal	74	65.6	Normal	74	65.5
		Mild	*22*	*19.5*	*Clinical levels*	*39*	*34.5*
		Moderate	*15*	*13.3*	*(Mild/Moderate*		
		Severe	*2*	*1.8*	*and Severe)*		

The above procedure was repeated for the regression analyses of positive and negative affect. In order to achieve approximately comparable groups, the sample was split in two using the mean score thus creating two groups; low and high levels of both positive and

negative affect. Table 5 shows the frequency and proportions of participants experiencing given levels of positive and negative affect on the *PANAS* scale.

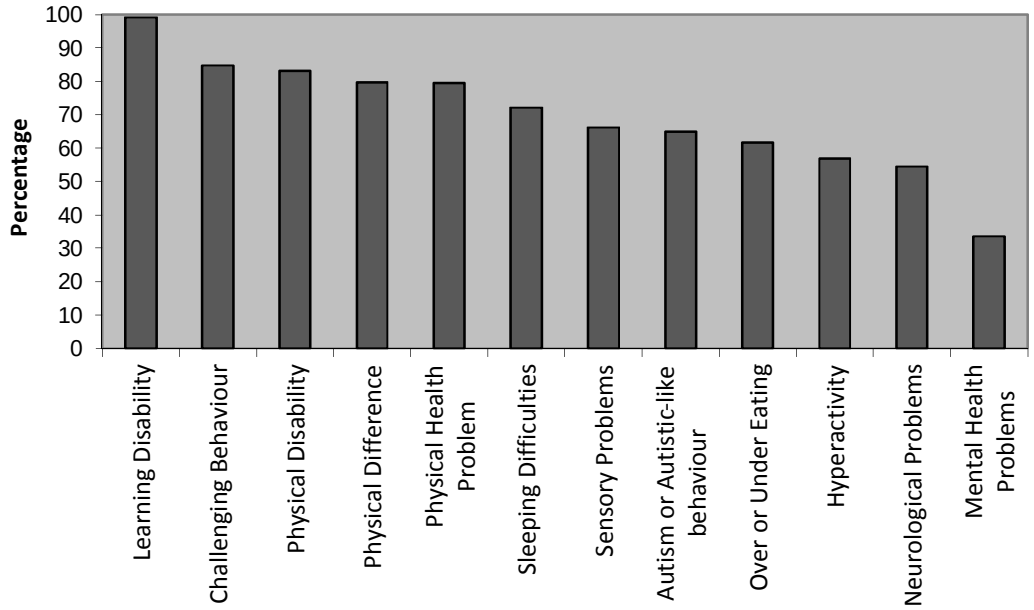
Table 5. Frequency of participants allocated to Low and High Positive Affect Groups

Affect	N	Category	N	Percentage
Positive	113	Low	59	52.2
		High	54	47.8
Negative	113	Low	63	55.8
		High	50	44.2

IPQ-RGS Identity Subscale Endorsement

As stated in the aims, in order to describe the levels of endorsement of child characteristics, percentages were calculated. Figure 2 shows the percentage of parents who endorsed each of the child characteristics contained within the *IPQ-RGS* subscales (see Appendix 6). The most frequently endorsed symptoms were learning disability (99.1%), challenging behaviour (84.7%) and physical disability (83%). With the exception of mental health problems (33%) all other characteristics (physical difference, physical health problems, neurological problems, sensory problems, autism or autistic like behaviour, sleeping difficulties, over or under eating and hyperactivity) were endorsed by at least 50% of the participants.

Figure 2. Parental Endorsement of *IPQ-RGS* Identity Subscale Items



Relationships between the *IPQ-RGS* Dimensions

Given that the *IPQ-RGS* was being used as a measure of illness representation in the current study, it was first necessary to assess the construct and discriminant validity of the *IPQ-RGS* subscales; therefore Pearson correlation coefficients were computed. This was necessary given that all further analyses will incorporate this measure. Inspection of the correlation matrix showed a number of significant inter-correlations. Table 6 shows the correlation coefficients of each of the *IPQ-RGS* dimensions.

Table 6. Inter-item correlations for subscales of the *IPQ-RGS*

	1	2	3	4	5	6	7
1. Identity							
2. Consequence	0.37**						
3. Personal Control	-0.06	-0.21					
4. Treatment Control	0.07	-0.16	0.50**				
5. Illness Coherence	0.16	0.21	-0.06	0.33			
6. Timeline Cyclical	0.24	0.10	0.05	0.11	-0.10		
7. Emotional representations	0.06	0.27**	-0.10	-0.05	-0.25**	0.15	

**p <0.01

The more a person attributes symptoms experienced to the genetic syndrome (illness identity) the stronger the perceptions of negative consequences of the genetic syndrome for the child (consequence). As beliefs about the negative consequences of the genetic syndrome on the child increased (consequences) stronger negative emotional representations are developed. Finally, as a persons' perception of their own ability to have a positive influence over the symptoms/characteristics of the genetic syndrome increased (personal control), so did the belief that the genetic syndrome is amenable to change (treatment control). Two significant negative associations were also observed. Stronger emotional representations were associated with poorer understanding of the genetic syndrome (illness coherence) and the stronger the belief that the genetic syndrome was impacting negatively on the child (consequence).

The Relationship between Illness Representations and Caregiver Wellbeing

In line with the study aims to provide a descriptive summary of the average scores on each of the measures by the current sample, means and standard deviations were calculated.

Table 7 details the means and standard deviations for *the COPE, HADS* and *PANAS*, age of child and level of disability.

Table 7. Mean Scores and Standard Deviations for the *IPQ-RGS, Brief COPE* Scales, the *HADS* and the *PANAS*

Scale	Subscale	Mean	SD
IPQ-RGS	Identity	8.31	2.29
	Consequence	26.31	3.32
	Personal Control	13.92	4.56
	Treatment Control	16.75	3.54
	Illness Coherence	20.15	3.42
	Timeline Cyclical	11.38	3.67
	Emotional Representation	19.58	5.26
Brief Cope	*Self Distraction*	*3.71*	*1.44*
	Active coping	*5.75*	*1.81*
	Denial	*2.25*	*0.84*
	Substance use	*2.85*	*1.51*
	Emotional support	*4.53*	*1.71*
	Instrumental support	*5.11*	*2.76*
	Behavioural disengagement	*2.43*	*0.91*
	Venting emotions	*3.5*	*1.35*
	Positive Reframing	*5.22*	*1.75*
	Planning	*5.83*	*1.80*
	Humour	*3.62*	*1.70*
	Acceptance	*6.93*	*1.41*
	Religion	*3.00*	*1.70*
	Self blame	*3.58*	*1.63*
HADS	Anxiety	9.75	4.72
	Depression	6.57	3.99
PANAS	*Positive affect*	*27.95*	*8.23*
	Negative affect	*18.13*	*7.77*
Demographics	Age of child	9.96	4.08
	Level of disability (Wessex score)	5.55	1.81

Relationship between IPQ-RGS Scales and Wellbeing Measures in Caregivers

In order to test the first hypothesis that participant's wellbeing may be related to illness perceptions and coping style, correlation co-efficients were calculated (see Appendix 11).

Table 8 shows the correlation coefficients for the *IPQ-RGS* dimensions, *HADS* and *PANAS* scales and age and level of disability of the child.

Table 8. Correlation coefficients for the *IPQ-RGS*, the *HADS* and the PANAS

	Anxiety	Depression	Positive Affect	Negative Affect	Age of Child	Level of Disability
Identity	-0.30*	-0.23	0.04	-0.20	0.14	0.06
Consequence	0.19	0.15	-0.04	0.18	0.02	-0.06
Personal Control	-0.09	-0.21	0.16	-0.13	-0.21	0.18
Treatment Control	-0.05	-0.04	0.15	-0.09	-0.12	0.09
Illness Coherence	-0.08	-0.25*	0.24	-0.09	-0.08	-0.04
Timeline Cyclical	0.26*	0.23	0.02	0.17	-0.08	-0.09
Emotional Representation	0.53*	0.41*	-0.19	0.52*	-0.23	-0.13

*p<0.01

Inspection of the correlation coefficients reveals that participants who reported higher levels of anxiety tended to have: stronger beliefs that the child's genetic syndrome is cyclical and unpredictable in nature (timeline cyclical), and held more negative emotional representations of the genetic syndrome. Furthermore participants who reported higher levels of anxiety tended to report fewer symptoms were associated with the genetic syndrome (identity). Depression is positively and significantly correlated to emotional representation and was negatively and significantly correlated illness coherence. Participants who reported higher levels of depression were less able to make sense of the genetic syndrome (illness coherence). Those participants who reported higher levels of depression also reported having more negative emotional representations of their child's genetic syndrome. Finally, participants who reported higher levels of negative affect also held more negative emotional representations of the genetic syndrome.

Relationship between the Brief COPE Scales and Wellbeing in Caregivers

In order to test the hypothesis that participant's wellbeing may be related to coping styles, correlation coefficients were calculated (see Appendix 11). Table 9 shows the correlation coefficients for the *brief COPE, HADS* and *PANAS* scales and age and level of disability of the child.

Table 9. Correlation coefficients for the *Brief COPE* Scales, the *HADS*, the *PANAS*, age of child and level of disability

	Anxiety	Depression	Positive Affect	Negative Affect	Age of Child	Level of disability
Self Distraction	0.14	0.13	0.08	0.26*	0.24	0.16
Active Coping	0.14	-0.01	0.39*	0.06	-0.56	0.08
Denial	0.09	0.20	-0.14	0.27*	-0.23	0.19
Substance Use	0.31*	0.33*	-0.13*	0.41*	0.16	0.17
Emotional Support	-0.06	-0.26*	0.25*	-0.02	-0.08	0.12
Instrumental Support	0.14	0.03	0.23	0.11	-0.10	0.08
Behavioural Disengagement	0.33*	0.36*	-0.35*	0.49*	0.04	0.16
Venting	0.21	0.17	0.08	0.33*	0.03	0.11
Positive Reframing	0.02	-0.04	0.41*	-0.03	-0.02	0.06
Planning	0.19	0.12	0.24*	0.20	0.04	0.35*
Humour	-0.02	-0.05	0.24	0.07	0.24	0.03
Acceptance	0.06	-0.04	0.32*	0.04	0.05	0.18
Religion	-0.06	-0.11	0.19	-0.02	0.05	-0.04
Self Blame	0.28*	0.39*	-0.14	0.46*	-0.08	0.17

*p<0.01

Inspection of the correlation coefficients revealed a number of both positive and negative significant associations. Those participants who reported higher levels of anxiety also report greater use of the coping strategies of substance use, behavioural disengagement, and self blame. Participants who reported higher levels of depression also reported greater use of the coping strategies of substance use, behavioural disengagement and self blame. In

addition to this they also reported less use of emotional support as a means to cope. Those participants who reported higher levels of positive affect reported using the following strategies to cope more frequently: active coping, emotional support, positive reframing, planning and acceptance. Those reporting higher levels of positive affect also endorsed the use of substances and behavioural disengagement as strategies to cope, less frequently. Those reporting higher levels of negative affect also endorsed the use of self distraction, substance use, emotional support, behavioural disengagement, venting and self blame as strategies to cope more frequently. Finally, the level of disability of the child was also positively and significantly related to the use of planning as a coping strategy, such that as level of disability of the child increases, so does the use of planning as a means to cope.

In summary, the analyses outlined above provide support for the hypothesis that anxiety, depression and positive affect are related to illness perceptions and coping styles. Anxiety, depression and positive affect were shown to have significant relationships with a number of the B*rief COPE* and *IPQ-RGS* subscales.

Relationship between Illness Representations and Coping Behaviours

Correlation coefficients were calculated in order to assess the second hypothesis that illness perception will be associated with coping behaviours (see Appendix 11). Table 10 shows the correlation coefficients for coping strategies and illness perceptions.

Table 10. Correlation coefficients for coping strategies and illness perceptions

	Identity	Consequence	Personal Control	Treatment Control	Illness Coherence	Timeline Cyclical	Emotional Representation
Self Distraction	-0.19	0.21	-0.14	-0.09	0.10	0.10	0.22
Active Coping	*-0.26**	*0.14*	*0.08*	*-0.16*	*0.10*	*0.02*	*0.10*
Denial	0.16	0.02	-0.03	-0.08	-0.32*	0.13	0.28*
Substance Use	*-0.21*	*0.11*	*-0.19*	*-0.05*	*0.06*	*0.09*	*0.25**
Emotional Support	-0.08	0.08	0.05	0.08	0.24	0.04	0.03
Instrumental Support	*-0.21*	*0.15*	*0.05*	*0.17*	*0.22*	*0.09*	*0.13*
Behavioural Disengagement	-0.11	0.11	-0.27	-0.29*	-0.05	-0.05	0.30*
Venting	*-0.34**	*0.20*	*-0.17*	*-0.04*	*0.08*	*0.14*	*0.29**
Positive Reframing	-0.02	0.06	0.19	0.11	0.08	0.10	-0.11
Planning	*-0.35**	*0.23*	*-0.03*	*0.09*	*0.08*	*-0.01*	*0.16*
Humour	-0.03	0.01	0.07	-0.01	0.01	0.01	-0.01
Acceptance	*-0.18*	*0.13*	*-0.02*	*-0.07*	*0.15*	*0.06*	*0.09*
Religion	0.04	0.03	0.01	0.08	-0.02	0.17	0.07
Self Blame	*-0.17*	*0.25**	*-0.09*	*-0.06*	*-0.003*	*0.15*	*0.41**

*p<0.01

Inspection of the correlation coefficients reveals a number of significant and positive correlations. Those who reported stronger beliefs that their child's genetic syndrome has negative consequences on their child's life reported higher levels of self blame. Those who

reported having stronger emotional representations reported higher levels of use of the following coping strategies: denial, substance use, behavioural disengagement, venting and self blame.

A number of significant negative correlations were also observed. Higher illness identity beliefs were associated with less use of the following coping strategies: active coping, venting and planning. Those who reported stronger beliefs that the symptoms of their child's genetic syndrome can be controlled through treatment also reported less use of the behavioural disengagement as a coping strategy. Finally participants who reported higher illness coherence also reported lower levels of denial. In summary, the analysis shows that hypothesis two was upheld in that illness perceptions were significantly related to coping behaviours being used by the participants in the current sample.

The Effect of Illness Representations on Caregiver Wellbeing

In order to examine the influence of illness perceptions and coping on anxiety, depression, positive and negative affect, four separate logistic regression analyses were conducted. Only those variables which correlated with anxiety, depression, positive and negative affect at $p < 0.01$ were entered into the model.

Predictors of Anxiety in Caregivers

In order to examine the influence of illness perceptions and coping on anxiety logistic regression analyses were conducted (see Appendix 12). Table 11 shows the logistic regression analysis of anxiety as a function of illness perceptions and coping. The analysis was performed on anxiety level as outcome with six predictors. Included in the analysis were: perceptions of illness identity, timeline cyclical, emotional representation and coping strategies of substance use, behavioural disengagement, and self blame.

Table 11. Logistic regression analyses of anxiety as a function of illness perception and coping

Variables	*B*	Wald Ratio (z-ratio)	Odds Ratio	95% Confidence Interval for Odds Ratio	
				Lower	Upper
Identity	0.17	1.88	1.19	0.93	1.51
Timeline Cyclical	0.15*	3.77	1.16	1.00	1.34
Emotional Representation	0.17**	7.56	1.18	1.05	1.34
Substance Use	0.73*	4.04	2.08	1.02	4.23
Behavioural disengagement	0.95	2.83	2.59	0.86	7.81
Self Blame	-0.10	0.30	0.91	0.63	1.29

R^2= 0.32 (Cox & Snell), *p<0.05, **p<0.01

The logistic regression model was significant (chi-squared =42.99, df = 6, p<0.01) and correctly classified 77.3% of cases. Three of the variables entered into model had significant dependent associations with anxiety; these were cyclical timeline, emotional representation, and the coping strategy of substance use.

Predictors of Depression in Caregivers

In order to examine the influence of illness perceptions and coping on depression logistic regression analyses were conducted. Table 12 shows the logistic regression analysis of depression as a function of illness perceptions and coping. The analysis was performed on depression level as outcome with six predictors. Included in the analysis were perceptions of illness coherence, emotional representation and coping strategies of substance use, emotional support, behavioural disengagement and self blame.

Table 12. Logistic regression analyses of Depression as a function of illness perception and coping

				95% Confidence Interval for Odds Ratio	
Variables	*B*	Wald Ratio (z-ratio)	Odds Ratio	Lower	Upper
Illness Coherence	-0.17*	5.01	0.84	0.72	0.98
Emotional Representation	0.09	2.86	1.09	0.99	1.21
Substance use	0.49**	6.27	1.63	1.11	2.39
Emotional Support	-0.35*	4.62	0.71	0.52	0.97
Behavioural disengagement	0.22	0.47	1.25	0.66	2.37
Self Blame	0.20	1.46	1.23	0.88	1.70

R^2= 0.32 (Cox & Snell) *p<0.05, **p<0.01

The logistic regression model was significant (chi-squared =36.54, df = 6, p<0.01) and correctly classified 77% of cases. Of the variables entered into model, three had a significant dependent association with depression, these being illness coherence and the coping strategies of substance use and emotional support.

Predictors of Positive Affect in Caregivers

In order to examine the influence of illness perceptions and coping on positive affect logistic regression analyses were conducted. Table 13 provides the details of the logistic regression analysis on positive affect as outcome with coping strategies of acceptance, substance use, emotional support, behavioural disengagement, positive reframing, planning and acceptance as predictors. Table 9 provides details of the regression analysis.

Table 13. Logistic regression analyses of positive affect as a function of illness perception and coping

Variables	*B*	Wald Ratio (z-ratio)	Odds Ratio	95% Confidence Interval for Odds Ratio	
				Lower	Upper
Active Coping	0.37*	5.08	1.44	1.05	1.98
Substance Use	-0.33	2.57	0.72	0.48	1.08
Emotional Support	0.20	1.55	1.22	0.89	1.67
Behavioural disengagement	-0.81*	4.37	0.44	0.21	0.95
Positive Reframing	0.35*	5.40	1.42	1.06	1.91
Planning	0.07	0.16	1.07	0.76	1.50
Acceptance	0.20	1.12	1.22	0.85	1.76

R^2= 0.34 (Cox & Snell), *p<0.05, **p<0.01

The logistic regression model was significant (chi-squared =46.17, df = 7, p<0.01) and correctly classified 74.3% of cases. Three of the variables entered into model had significant dependent associations with positive affect these were active coping, behavioural disengagement and positive reframing.

Predictors of Negative Affect in Caregivers

In order to examine the influence of illness perceptions and coping on negative affect logistic regression analyses were conducted. Table 14 provides the details of the logistic regression analysis on negative affect as outcome with seven predictors. Included in the analysis were emotional representation and the coping strategies of self distraction, denial, substance use, behavioural disengagement, venting and self blame. Table 9 provides details of the regression analysis.

Table 14. Logistic regression analyses of negative affect as a function of illness perception and coping

Variables	*B*	Wald Ratio (z-ratio)	Odds Ratio	95% Confidence Interval for Odds Ratio	
				Lower	Upper
Emotional Representation	0.15*	7.79	1.16	1.05	1.29
Self Distraction	0.10	0.20	1.11	0.71	1.74
Denial	0.13	0.09	1.14	0.50	2.57
Substance Use	0.31	2.73	1.36	0.94	1.97
Behavioural Disengagement	0.55	1.91	1.73	0.80	3.75
Venting	0.30	1.48	1.36	0.83	2.21
Self Blame	0.10	0.45	1.11	0.82	1.50

R^2= 0.25 (Cox & Snell), *p<0.05, **p<0.01

The logistic regression model was significant (chi-squared =39.62, df = 7, $p<0.01$) and correctly classified 72.6% of cases. One of the variables entered into model had a significant dependent association with negative affect this being emotional representation.

Discussion

This study is the first to examine the use of a modified version of the *IPQ-R* to investigate illness perceptions in the caregivers of children with rare genetic syndromes. The psychometric properties of the *IPQ-RGS* were examined along with the associations between illness perceptions and coping styles. Psychological wellbeing outcomes for the caregivers were also investigated. Previous studies examining illness perceptions in relation to outcomes have generally focused on psychological distress. In the current study psychological distress was examined (anxiety, depression and negative affect) and a measure of positive affect was also taken to take into account the positive experiences of parenting a child with a genetic syndrome (Horsley and Oliver, in press).

The results suggest that psychological outcomes for parents are related to illness perceptions and coping strategies, with a number of significant relationships observed between illness representations, coping and psychological outcomes. Regression analyses revealed a number of relationships, predictable based on the common sense model of illness representations, between illness perceptions, coping and psychological wellbeing providing preliminary support for the hypothesis that the common sense model of illness representations can be applied to the caregivers of children with rare genetic syndromes.

A number of modifications were made to the *IPQ-R* resulting in the development of the *IPQ-RGS*. The timeline acute/chronic scale was omitted as it was deemed inappropriate given that caregivers of children with rare genetic syndromes will be aware the genetic syndrome is not something that will be temporary in nature. Furthermore, the identity subscale contains a list of generic items; participants are asked to indicate which symptoms/features that their child has experienced and are then asked to indicate if they feel that this particular symptom/feature is related to the genetic syndrome. It is the sum of the second of these that provides a score of illness identity. Given that no such scale had previously been developed, one was created for the purpose of the current study. Analysis revealed that the identity subscale designed for the *IPQ-RGS* does provide a measure of illness identity rather than simply measuring somatisation. Due to the fact that the characteristics within the identity subscale will be more symptomatic of some genetic syndromes compared to others, internal reliability of this scale is less important (see Moss-Morris et al., 2002).

The construct validity of the *IPQ-RGS* was also examined; it was hypothesised that the *IPQ-RGS* dimensions would correlate in a pattern similar to that observed in previous studies. This hypothesis was also supported. For example Al Anbar et al., (2010) adapted the *IPQ-R* for use with parents of children with autism spectrum disorders (*IPQ-RA*). The following inter-correlation similarities were observed between the current findings and those observed in the *IPQ-RA*: Consequence with identity (positive correlation), consequence with emotional representations (positive correlation) and illness coherence and emotional representations (negative correlation). Furthermore, the inter-correlations observed within

the current study partially replicate the meta-analysis findings of Hagger and Orbell (2003). The similarities observed were the positive and significant correlation coefficients for the identity-consequences and identity-timeline cyclical dimensions, and also the negative and significant correlation coefficient for personal control-consequences dimensions. This also indicates that the common sense model of illness representations can be applied reliably to the caregivers of children with rare genetic syndromes.

In summary, the *IPQ-RGS* has been shown to be an effective measure of illness perceptions demonstrating good internal consistency and can thus be used to assess caregiver illness perceptions within this population. The reliability of the individual scales of the *IPQ-RGS* are comparable to those found by a number of authors who have adapted the scale in similar ways (Al Anbar et al., 2010; Barrowclough et al., 2005; Fortune et al., 2001; Lobban et al., 2001).

A number of significant relationships were observed between the dimensions of illness representation, coping and psychological wellbeing which were consistent with previous research findings. These relationships were predictable based upon Leventhal et al.'s (1980) common sense model, suggesting that illness representations may have important implications for coping strategies and caregiver wellbeing. All regression models were significant statistically and correctly classified between 70 and 77.3% of cases, suggesting that knowledge of aspects of person's illness perception and the coping strategy being adopted, can predict psychological wellbeing.

Leventhal et al., (1997) suggest that illness perceptions are developed in a way which helps to maintain internal coherence, which suggests that whilst illness perceptions are susceptible to change, they are a more schematic representation that remains somewhat stable over time. The pattern of inter-correlations observed within the current study demonstrates that the cognitive and emotional representations assessed for caregivers have good internal validity suggesting the illness representations held by the caregivers of children in the current study are likely to represent a stable schematic representation. This finding also reflects previous research findings, including those of Barrowclough et al. (2001), Fortune et al. (2005), Al Anbar et al. (2010), Hagger and Orbell (2003) and Williams and Rose (2007).

The results suggest that caregivers who have strong beliefs that their child's genetic syndrome is cyclical and unpredictable in nature, also tend to develop a stronger emotional representation. These caregivers are more likely to use substances and are more likely to report clinical levels of anxiety. It was not possible to establish a causal direction within the current study given the correlational methodology. Illness coherence, substance use and emotional support were also found to have significant dependent associations with depression. In terms of the common sense model of illness representations, it could be hypothesised that those caregivers who are unable to make sense of and create a coherent understanding of their child's genetic syndrome are more likely to use substances as a means to cope, are less likely to seek emotional support and are at an increased risk of

experiencing clinical levels of depression. Further research would be needed to establish the direction of relationships.

The above findings are important as they provide evidence to support the common sense model as a theoretical model for use within the current population. In summary, these findings, coupled with the theoretically predictable relations from the common sense model of illness representations, suggest that in order to improve outcomes for the caregivers of children with genetic syndromes, interventions should be designed to target the perceptions that a caregiver holds about their child's genetic syndrome. Interventions could also specifically target the use of coping strategies, helping caregivers to develop more helpful ways of coping and reducing the use of strategies considered less helpful.

In the current study caregivers with a poor understanding of the genetic syndrome and those who hold beliefs that the symptoms/features of the genetic syndrome will be cyclical and unpredictable in nature, are likely to experience higher levels of psychological distress. Therefore, in order to improve the emotional wellbeing of caregivers, health professionals should be encouraged to help caregivers understand the genetic syndrome helping them to develop a clear and coherent picture of it. It may be the case that certain symptoms/characteristics of a genetic syndrome are unpredictable and so when caregivers have strong beliefs in relation to this, then interventions may need to target caregiver coping styles. Furthermore, whilst illness coherence was not included in the regression model for positive affect (due to it not reaching significant to 0.01) inspection of the correlation coefficients did reveal a significant positive relation to illness coherence, suggesting that

those caregivers who had a clearer understanding of the genetic syndrome were more likely to experience positive emotions again suggesting that by improving a caregiver's understanding of the genetic syndrome will have a positive impact on psychological outcomes.

This is the first study of this nature to include a measure of positive outcome. Findings revealed that caregivers reporting higher levels of positive affect were more likely to report engaging in positive reframing and active coping as a means to cope and were significantly less likely to report engaging in behavioural disengagement. Once again however, it is important to hold in mind the limitations of the current study in that the statistical analysis does not allow causal statements to be made. These findings provide only partial support for the common sense model as no statistically significant relationships were found between positive affect and illness representation dimensions. One explanation for this finding may be that the measure of positive affect describes a mood state over the previous fortnight and can be considered a measure of state positive affect whereas illness representations, whilst changeable, are considered more stable (more trait like). As such, it may be more appropriate in future studies to assess positive outcomes of a more stable nature such as positive gains and family hardiness as these can be considered more stable measures of positive outcomes. In this way, the common sense model of illness representations is also able to capture and explain the differences observed between studies where some have described caregivers experiencing depression when other studies have found no such effects.

There are a number of methodological and statistical weaknesses of the current study which must be borne in mind when interpreting the findings. Firstly, the sample is biased due to recruitment from a pool of participants who have previously been involved in related research. The majority of the participants in the current study will have been recruited through support groups; there is no way of knowing if these caregivers are comparable to those who do not access the support groups. In the current study this is particularly important given that at least some point in the past the current participants will have taken an active approach to coping and sought support which may in some way have skewed the findings. Given that the response rate for the current study was also low (29.2%) this also brings into the question the motivation for the current sample to take part in this study. One way to circumnavigate this issue would be to recruit from a wider range of caregivers, perhaps through media advertising, and/or recruiting through other services including social services, schools and healthcare providers. Furthermore, the current study combines the responses of caregivers, regardless of the genetic syndrome that their child has been diagnosed with. It may be worthwhile considering repeating the current study with a larger sample from each of the genetic syndrome groups as this would allow for an assessment of between group differences. It may be that a particular group of parents is more or less likely to experience positive outcomes or anxiety or depression and it may be that specific patterns of illness perceptions is observed between groups.

The findings within the current study fit into a broader theoretical framework. They provide a step forward in our understanding of how caregivers perceive their child's genetic

syndrome and suggest that the common sense model of illness representations can be applied to this group of individuals. They also suggest that in line with previous research, it would be helpful to understand all manner of positive outcomes for caregivers. Given that it has been well established within the physical health domain that illness representations will impact upon help seeking behaviours, it is now important to explore the common sense model within this sample whilst also incorporating a wider range of outcome measures including outcomes for the children for whom they care.

References

Al Anbar, N.N., Derdennes, R.M., Prado-Netto, A., Kaye, K. & Contejan, Y. (2010) Treatment choices in autism spectrum disorder: The role of parental illness perceptions. *Research in Developmental Disabilities, **31***, 817-828.

Arron, K., Oliver, C., Berg, K., Moss, J. and Burbidge, C. (2011). Prevalence and phenomenology of self-injurious and aggressive behaviour in genetic syndromes. *Journal of Intellectual Disability Research*, ***55***, 109-120.

Barrowclough, C., Lobban, F., Hatton, C. & Quinn, J. (2001). An investigation of models of illness in carers of schizophrenia patients using the illness perception questionnaire. *British Journal of Clinical Psychology, **40***, 371-385.

Baron, R.M. & Kenny, D.A. (1986) The moderator-mediator variable distinction in social psychological research: Conceptual, strategic and statistical considerations. *Journal of Personality and Social Psychology, **51***, 1173-1182.

Berg, K., Arron, A., Burbidge, C., Moss, J. & Oliver, C. (2007). Carer-reported contemporary health problems in people with severe and profound intellectual disability and genetic syndromes. *Journal of Policy Practice in Intellectual Disabilities, **4***, 120-128.

Bull, L., Woodcock, K.A., Perry, A., Todd, H., Holland, A.J. & Oliver, C. (2011) Temper outbursts in Prader-Willi syndrome: Early intervention and environmental management. *Journal of Intellectual Disability Research, **55**,* 109-120.

Burbidge, C., Oliver, C., Moss, J., Arron, K., Berg, K., Hill, L., Trusler, K., Furniss, F. and Woodcock, K. A. (2010). The association between repetitive behaviours, impulsivity and hyperactivity in people with intellectual disability. *Journal of Intellectual Disability Research,* 54, 1078-1092.

Carver, C.S. (1997) You want to measure coping but your protocol's too long: Consider the brief COPE. *International Journal of Behavioral Medicine, **41***, 92-100.

Carver, C.S., Scheier, M.F. & Weintraub, J.K. (1989). Assessing coping strategies: A theoretically based approach. *Journal of Personality and Social Psychology, **56**,* 267-283.

Department of Health (2008). *Carers at the heart of the 21st century*. London: DoH

DeVellis, R.F. (1991) *Scale development.* Newbury Park, CA: Sage *cited by* Al Anbar, N.N., Derdennes, R.M., Prado-Netto, A., Kaye, K. & Contejan, Y. (2010) Treatment choices in autism spectrum disorder: The role of parental illness perceptions. *Research in Developmental Disabilities, **31***, 817-828.

Dura-Vila, G., Dein, S. & Hodes, M. (2010) Children with intellectual disability: A gain not a loss: Parental beliefs and family life. *Clinical Child Psychology, **15***, 171-184.

Emerson, E., Roberston, J. & Wood, J. (2004) Levels of psychological distress experienced by family carers of children and adolescents with intellectual disabilities in an urban conurbation. *Journal of Applied Research in Intellectual Disability, **17***, 77-84.

Fortune, D.G., Smith, J.V. & Garvey, K. (2005) Perceptions of psychosis, coping, appraisals and psychological distress in the relatives of patients with schizophrenia: An exploration using self-regulation theory. *British Journal of Clinical Psychology, **44***, 319-331.

Foster, R. H., Kozacehk, S., Stern, M. & Elsea, S.H. (2010) Caring for the caregivers: An investigation of factors related to well-being among parents caring for a child with smith-magenis syndrome. *Journal of Genetic Counselling, **19**,* 187-198.

Griffith, G.M., Hastings, R.P., Oliver, C., Howlin, P., Moss, J., Petty, J., & Tunnicliffe. P. (2011) Psychological well-being in parents of children with angelman, cornelia de lane and cri du chat syndromes. *Journal of Intellectual Disability Research, **44***, 397-410.

Hagger, M.S. & Orbell, S. (2003) A meta-analytic review of the common-sense model of illness representations. *Psychology and Health, **18***, 141-184.

Hassall, R. & Rose, J. (2005) Parental cognitions and adaptation to the demands of caring for a child with an intellectual disability: A review of the literature and implications for clinical interventions. *Behavioural and Cognitive Psychotherapy, **33,** 71-88.*

Hassall, R., Rose, J. and MacDonald, J. (2005). Parenting stress in mothers of children with an intellectual disability: the effects of parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research, **49,** 405-418*

Hastings, R. & Taunt, H. (2002) Positive perceptions in families of children with intellectual disabilities. *American Journal of Mental Retardation, **107***, 116-127

Hastings, R.P., Beck, A. & Hill, C. (2005). Positive contributions made by children with an intellectual disability in the family: mothers and fathers perspectives. *Journal of Intellectual Disabilities, **9***, 155-165.

Hastings, R. P., Daley, D., Burns, C., & Beck, A. (2006). Maternal distress and expressed emotion: Cross-sectional and longitudinal relationships with behavior problems of children with intellectual disabilities. *Journal Information*, ***111*****,** 48-61.

Hayes, S., McGuire, B., O'Neil, M., Oliver, C., & Morrison, T. (2011) Low mood and challenging behaviour in people with severe and profound intellectual disabilities. *Journal of Intellectual Disability Research, **55***, 182-189.

Herring, S., Gray, K., Taffe, J, Tonge, B, Sweeney, D. & Einfeld, S. (2006) Behaviour and emotional problems in toddlers with pervasive developmental disorders and developmental delay: associations with parental mental health and family functioning. *Journal of Intellectual Disability Research, **50***, 874-872.

Hill, C. & Rose, J. (2009) Parenting stress in mothers of adults with an intellectual disability: parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research, **53***, 969-980.

Horsley, S. and Oliver, C. (in press) Positive impact and its relationship to wellbeing in parents of children with intellectual disability: a literature review. Submitted to *British Journal of Developmental Disorders.*

Hyman, P. and Oliver, C. (2001) Causal explanations, concern and optimism regarding self-injurious behaviour displayed by individuals with cornelia de lange syndrome: The parents perspective. *Journal of Intellectual Disability Research**, 45***, 326-344.

Johnstone, C., Hessl, D., Blasey, C., Eliez., S., Erba, H., Dyer-Friedman, J., Glaser, B. & Reiss, A.L. (2010). Factors associated with parenting stress in mothers of children with fragile X syndrome. *Developmental and Behavioral Paediatrics,* ***24,*** 267-275.

Kim, H.W., Greenberg, J.S., Seltzer, M.M., & Krauss, M.W. (2003). The role of coping in maintaining the psychological wellbeing of mothers of adults with intellectual disability and mental illness. *Journal of Intellectual Disability Research,* ***47,*** 313-327.

Kushlick, A., Blunden, R. & Cox, G. (1973) A method of rating behaviour characteristics for use in large scale surveys of mental handicap. *Psychological Medicine,* ***3***, 466-478.

Leventhal, H., Meyer, D & Nerenz, D. (1980). The common sense model of illness danger. In S. Rachman (Ed.) *Medical Psychology*, Vol 2. pp 7-30. Pergamon, New York.

Leventhal, H., Benyamini, Y., Brownlee, S., Diefenbach, M., Leventhal, E., Patrick-Miller, L. & Robitaille, C. (1997). Illness representations: Theoretical foundations. In: K. J. Petrie, & J. Weinman (Eds.), *Perceptions of health and illness: Current research and applications* (pp. 19–45). Amsterdam: Harwood Academic Publishers.

Lobban, F. Barrowclough, C. and Jones, S. (2003) A review of the role of illness models in severe mental illness. *Clinical Psychology Review,* ***23,*** 171-196.

Meyer, D., Leventhal, H. & Gutmann, M. (1985). Common-sense models of illness: the example of hypertension. *Health Psychology,* ***4*****,** 115-135.

Miller, A.C., Gordon, R.M., Daniele, R.J. and Diller, L. (1992). Sress, appraisal, and coping in mothers of disabled and non disabled children. *Journal of Paediatric Psychology,* ***17,*** 587-605.

Moss, J., Oliver, C., Arron, K., Burbidge, C. and Berg, K. (2009). The prevalence and phenomenology of repetitive behavior in genetic syndromes. *Journal of Autism and Developmental Disorders,* ***39,*** 572-588.

Moss-Morris, R., Weinman, J., Petrie, K.J., Horne, R, Cameron, L.D. & Buick, D. (2002) The revised illness perception questionnaire (IPQ-R). *Psychology and Health,* ***17*****,** 1-16.

Norizan, A. and Shamsuddin, K. (2010) Predictors of parenting stress among Malaysian mothers of children with down syndrome. *Journal of Intellectual Disability Research,* ***54***, 992- 1003.

Oliver, C., Arron, K., Sloneem, J., & Hall, S. (2008). Behavioural phenotype of Cornelia de Lange syndrome: case–control study. *The British Journal of Psychiatry*, *193*:6, 466-470.

Oliver, C. & Woodcock, K. (2008). Integrating levels of explanation in behavioural phenotype research. *Journal of Intellectual Disability Research,* ***52***, 807-809.

Oliver, C., Woodcock, K. & Adams, D. (2010). The importance of aetiology of intellectual disability. In: Grant, G., Ramcharan, P., Flynn, M. and Richardson, M. (Eds.) Learning Disability: A life cycle approach to valuing people. (pp. 135-146). Open University Press:Wiley.

Orsmond, G. I., Seltzer, M. M., Krauss, M. W., & Hong, J. (2003). Behavior problems in adults with mental retardation and maternal well-being: Examination of the direction of effects. *Journal Information*, ***108*,** 53-66.

Padeliadu, S. (1998) Time demands and experienced stress in Greek mothers of children with down's syndrome. *Journal of Intellectual Disability Research, **42***, 144-153.

Singer, G.H.S. (2006) Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation, **111***, 155-169.

Sloneem, J., Oliver, C., Udwin, O., & Woodcock, K.A. (2011) Prevelace, phenomenology, aetiology and predictors of challenging behaviour in smith-magenis syndrome. *Journal of Intellectual Disability Research, **55***, 138-151.

Teixeira, M.C.T.V., Emerich, D.R., Orsati, F.T., Rimerio, R.C., Gatto, K.R., Chappaz, I.O. & Kim C.A. (2011) A description of adaptive and maladaptive behaviour in children and

adolescents with cri-du-chat syndrome. *Journal of Intellectual Disability Research,* ***55***, 132-137.

Van den Borne, H.W., van Hooren, R.H., van Gestel, M., Rienmeijer, P., Fryns, J.P., & Curfs., L.M.G. (1999). Psychosocial problems, coping strategies and the need for information of parents of children with prader-willi syndrome and angelman syndrome. *Patient Education and Counselling,* ***38***, 205-216.

Watson, D., Clark, L.A. & Tellegan, A. (1988) Development and validation of brief measures of positive and negative affect: The PANAS scale. *Journal of Personality and Social Psychology,* ***54*****,** 1063-1070

Weiner, B. (1985) An attributional theory of achievement motivation and emotion. *Psychology Review,* ***92****,* 548-573.

Weiner, B. (1980) A cognitive (attribution)-emotion-action- model of motivated behaviour: an analysis of judgements of help-giving. *Journal of Personality and Social Psychology,* ***39***, 186-200.

Weinman, J., Petrie, K.J., Moss-Morris, R. and Horne, R. (1996) The illness perceptions questionnaire: A new method for assessing the cognitive representation of illness. *Psychology and Health,* ***11***, 43105-43114.

Williams, R.J. & Rose, J.L. (2007) The development of a questionnaire to assess the perceptions of care staff towards people with intellectual disabilities who display challenging behaviour. *Journal of Intellectual Disabilities, **11***, 197-211.

Wulffaert, J., van Berckelaer-Onnes, I., Kroonenberg, P., Scholte, E. & Hennekam, R. (2009) Simultaneous analysis of the behavioural phenotype, physical factors and parenting stress in people with cornelia de lange syndrome. *Journal of Intellectual Disability Research, **53***, 604-619.

Zigmond, A.S. and Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica, 67*, 361-370.

Public Domain Briefing Paper

Parental Perceptions of Children's Genetic Syndromes and their Impact on Coping and Mental Health

~ Natalie Byrne ~

University of Birmingham

Background and Aims

Before this study was conducted, previous studies had told us that the parents of children with rare genetic syndromes sometimes experience common mental health problems such as depression and anxiety (Griffith et al., 2011; Horsley and Oliver, In press; Norizan and Shamsuddin, 2010). However, it is important to recognise that parents of children with intellectual disability also have many positives experiences of caring for their child (Horsley and Oliver, in press). They suggest that these positive experiences may reduce negative outcomes for these parents. It is known that anxiety and depression are influenced by cognitive factors, or put simply, the way that parents think about things. It is therefore important to understand which factors affect the way that parents think about their children and specifically their genetic syndromes. Understanding this might help us to understand the way that parents cope with caring for their child and also the way that this impacts on their mental health.

An empirical study was conducted to explore the impact of the way that a parent thinks about their child's genetic syndrome, on the way that they cope, how they feel and their mental health. To capture the way that a parent thinks about their child's genetic syndrome, also known as their perception of it, a framework called the Common Sense Model of Illness Representations was used (Leventhal et al., 1980). This framework captures many different aspects of parent's perceptions about their child's genetic syndrome.

It was predicted that the Commons Sense Model of Illness Representations would be a useful way of understanding how parents perceive their child's genetic syndrome and how this will impact upon the behaviours that they choose to cope with difficulties, their feelings generally and also their mental health. It was predicted that there would be relationships between different parts of the model and between these and coping behaviours and mental health and that these would be similar to the patterns observed in other groups of individuals where it has been used.

Methodology

In the empirical study 113 parents completed a number of questionnaires. One questionnaire, The Illness Perceptions Questionnaire- Revised for Genetic Syndromes

(designed for the purpose of the current study) captured parent's perceptions of their child's genetic syndrome, one captured their current mood (The Positive and Negative Affect Scale, Watson et al., 1988), another asked about their coping behaviours (The Brief COPE, Carver et al., 1997) and the other was an assessment of their mental health (asking about anxiety and depression; Hospital Anxiety and Depression Scale, Zigmond and Snaith, 1983). Perceptions included the parent's ideas about the label attached to their child's genetic syndrome, the parent's perception of the likely duration of the illness and its symptoms, the person's perception of severity of the illness and the impact on areas of their functioning including physical, social and psychological and the person's perceptions of the degree to which the illness can be controlled and/or cured by treatments or by actions of the individual and how the illness "makes sense as a whole to the patient".

Findings

The study findings were in line with the predictions made. A number of relationships were observed between different aspects of parents perceptions and these could reliably predict parents coping behaviours, feelings and anxiety and depression. This suggests that the Common Sense Model of Illness Representations is a useful framework for understanding the way that parents think about their child's genetic syndrome. The relationships were also

similar to those observed in previous studies in different groups of individuals (Hagger and Orbell, 2003). The findings also showed that a number of parent's were experiencing some levels of anxiety and/or depression.

Importance of the Findings

The current study showed that some parents of children with rare genetic syndromes are experiencing anxiety and depression; this suggests that more needs to be done to support parents and alleviate their distress. Findings also suggest that the way that parents think about their child's genetic syndrome is related to the way that they cope and feel. In the current study caregivers with a poor understanding of the genetic syndrome and those who hold beliefs that the symptoms/features of the genetic syndrome will be cyclical and unpredictable in nature, are likely to experience higher levels of psychological distress. Therefore, in order to improve the emotional wellbeing of caregivers, health professionals should be encouraged to help caregivers understand the genetic syndrome helping them to develop a clear and coherent picture of it. It may be the case that certain symptoms/characteristics of a genetic syndrome are unpredictable and so when caregivers have strong beliefs in relation to this, then interventions may need to target caregiver coping

Future Implications

The findings provide a step forward in our understanding of how parents perceive their child's genetic syndrome and suggest that the common sense model can be applied to this group of individuals. It would be helpful to understand all manner of positive outcomes for parents also. Given that it has been well established within the physical health research that perceptions of an illness will impact upon help seeking behaviours, it is now important to explore the common sense model with parents of children with genetic syndromes, whilst also including a wider range of outcome measures including outcomes for the children for whom they care.

References

Carver, C.S. (1997) You want to measure coping but your protocol's too long: Consider the brief COPE. *International Journal of Behavioral Medicine, **41***, 92-100.

Griffith, G.M., Hastings, R.P., Oliver, C., Howlin, P., Moss, J., Petty, J., & Tunnicliffe. P. (2011) Psychological well-being in parents of children with angelman, cornelia de lane and cri du chat syndromes. *Journal of Intellectual Disability Research, **44***, 397-410.

Hagger, M.S. & Orbell, S. (2003) A meta-analytic review of the common-sense model of illness representations. *Psychology and Health, **18***, 141-184.

Horsley, S. and Oliver, C. (in press) Positive impact and its relationship to wellbeing in parents of children with intellectual disability: a literature review. Submitted to *British Journal of Developmental Disorders.*

Leventhal, H., Meyer, D & Nerenz, D. (1980). The common sense model of illness danger. In S. Rachman (Ed.) *Medical Psychology*, Vol 2. pp 7-30. Pergamon, New York.

Norizan, A. and Shamsuddin, K. (2010) Predictors of parenting stress among Malaysian mothers of children with down syndrome. *Journal of Intellectual Disability Research, **54***, 992- 1003.

Watson, D., Clark, L.A. & Tellegan, A. (1988) Development and validation of brief measures of positive and negative affect: The PANAS scale. *Journal of Personality and Social Psychology,* ***54*****,** 1063-1070

Zigmond, A.S. and Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica, 67*, 361-370.

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Appendices

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Appendix 1- Information Sheet for Participants

UNIVERSITY OF BIRMINGHAM

Parental understandings and feelings about Childs Genetic Syndrome, Challenging Behaviour and its Impact upon Parental and Child Wellbeing and Service Provision:

Information Sheet

Please read this information carefully before deciding whether you wish to take part in the study. If you have any further questions please contact with Natalie Jackson at NEJ909@adf.bham.ac.uk, Efthalia Karakatsani on 01214142855 or at exk085@bham.ac.uk. If you have any medical/ other problems which make it difficult for you to read this information, please contact with Natalie Jackson or Efthalia Karakatsani for a verbal explanation of the research.

When you are happy that you have all of the information you need to be able to decide whether or not you and the person you care for would like to take part in the study, please complete the enclosed consent form and questionnaire pack return them to us in the prepaid envelope provided

Background

We would like to invite you to take part in a questionnaire study being conducted at the Centre for Neurodevelopmental Disorders, University of Birmingham. This research work, which is led by Professor Chris Oliver, looks at parental understandings and feelings about Childs Genetic Syndrome and Challenging Behaviour We hope that this information will enable us to define the association of challenging behaviour with parental perspectives of child's disability and family wellbeing with service provision.

Aims of the study

This study aims to further our understanding of challenging behaviour in individuals with intellectual disabilities. Eventually we hope that our results will help to improve the quality of life of individuals with intellectual disabilities and their families.

What will happen if you and your child/the person you care for decide(s) to participate?

Where will the research take place?

The research will involve completing the enclosed questionnaire pack. This can be completed by you in your own time at your home.

Who will be involved in collecting the data?

Members of the research team at the Cerebra Centre for Neurodevelopmental disorders including Natalie Jackson and Efthalia Karakatsani.

How long will participation in the study take?

The questionnaire pack will take approximately 45 minutes to complete.

In the future you may be asked if you would like to complete the questionnaire again so that we can start to understand what happens to people with intellectual disabilities across their lifetime. We will only contact you with this invitation if you have previously agreed to be contacted by the research team at the University of Birmingham with information about research studies conducted by the team.

We will be collecting information from participants between September 2012 and March 2015. After this we will spend some time understanding the information we have collected and writing reports. This means that the study will be finished in 18 months after end date of data collection phase.

Sometimes after you have completed the questionnaire, we may need to contact you again in order to clarify any information that you have provided or to ask you for further information regarding the diagnosis of the person you care for. This helps us to ensure that our data is as useful and as accurate as possible. If this happens then we would contact you again within 6 months of receiving your questionnaire pack to ask whether or not you would be willing to provide us with the extra information.

What will participants are required to do during the study?

You will be asked to take part in an online survey that will be conducted by researchers online. We would like to complete some standardized questionnaires about the behaviour of your child/person you care for. The questionnaires will take approximately 45 minutes.

Are there any risks that individuals taking part in the study might face?

There will not be any risks associated with participation in this study.

What are the potential benefits for participants from taking part?

You will receive personalised feedback regarding your child/ the person you care for. This study will help us to find out more about the lives of people with challenging behaviour and the difficulties that these people face. It will also help us to understand your beliefs about this. The results might help us to improve things for people with challenging behaviour in the future.

Where will data be stored?

The data collected will be kept in locked or password protected storage at the University of Birmingham. All information gathered about you and your child will be stored separately from any information that would allow someone to identify who you or your child are (this is known as personal identifying information, e.g. your full names, your address, your contact details). Your personal identifying information will be stored in a locked space at the University of Birmingham and only members of our research team will have access to it. We will only be able to trace the information we have collected about you and your child back to you using a special reference number which we will store in a password protected database held at the University of Birmingham. Only members of our research team will have access to that database. Personal identifying information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998.

The video and/or audio recordings are considered to necessarily contain personal identifying information. We will therefore store the recordings of you and your child separately to the other information we have collected about you. These recordings will not be labelled with your names or any other personal identifying information but will be labelled with your special reference number. Recordings will be stored in a locked cabinet at the University of Birmingham and only members of our research team will have access to it.

If you/ the person you care for decide(s) to participate, what will happen after that participation?

You and your child/ person you care for will receive an individual feedback report describing the results of all of the assessments that were carried out during the study. If requested, this feedback report will be circulated to other interested individuals. Descriptions of research findings will be published in newsletters of the relevant family support groups and educational institutions involved. Any request for advice concerning the person you care for will be referred to Professor Chris Oliver, Clinical Psychologist.
The researchers will publish the findings from the study in scientific journals and will present the results at relevant conferences.

What will happen to the data afterwards?

The information that you provide will be locked in a filing cabinet at the University of Birmingham or held on a password protected database. Participants will be identified by a unique number so that the information you provide us with cannot be traced to your personal details. You will be able to decide whether or not you want to make your research data available to any professionals or clinicians working with you and the person you care for should they wish to see it. This is optional and will not affect your participation in the current study. If you agree to this, then your research data will only be made available to relevant clinicians or professionals should they contact us directly and request to see it. If you do not agree to this then research data will not be made available to anyone other than the research team at the University of Birmingham.

After 6 months of receiving your questionnaire, your personal details will be ***destroyed unless you tell us otherwise.*** This means that we would no longer be able to trace the results of your assessments back to you. ***The section below on 'The Regular Participant Database Information'*** gives information about a database that we use to store the personal details of some participants. Please read this section in order to decide if you would like to join that database.

Regular Participant Database Information:

What is the regular participant database?

We have a database that we keep in the Cerebra Centre where we store the names and contact details of some previous participants. If you would like us to, we can add your details to this database. We would use this information for two things:

1) We will contact you with information about future research work to find out whether or not you would like to participate.
2) It is often important to find out how things change over time. By keeping your details we would be able to trace the results of the previous assessments that you have done with us back to you. This means that if you take part in other studies with us we would be able to look at how things have changed over time.

Who would have access to my details?

Only approved members of our research team would have access to your details. We would not share your details with anyone outside the research team.

When would I be contacted?

You would only be contacted by an approved member of the research team when we are starting another study or phase of a study that we think you might like to participate in or when we need to clarify some information that you have provided us with from participation in a research study.

What happens if I decide that I want my details to be added to the database but then I change my mind?

All you would need to do is contact Chris Oliver on 0121 414 7206 or at c.oliver@bham.ac.uk or at the School of Psychology, University of Birmingham, Edgbaston, Birmingham, B15 2TT. Your details would be removed from the database immediately.

Consent

After having read all of the information and having received appropriate responses to any questions that you may have about the study you and the person you care for will be asked to give you and your child's/ person you care for's consent to participate in the study if you decide that you do wish to participate. The section below on ***'Giving consent'*** will explain this process. We need to receive consent from/ on behalf of potential participants in order for them to participate.

Withdrawal

Even after consent has been granted, participants can request to be withdrawn from the study at any time, without giving a reason. Even after participation has taken place, consent can be withdrawn and any data collected will be destroyed. This will not restrict the access of you/ the person you care for to other services and will not affect their right to treatment.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. Please contact Chris Oliver on 0121 414 7206 or at c.oliver@bham.ac.uk in the first instance. If you remain unhappy and wish to complain formally, you can contact: Professor Chris Miall; Head of School; School of Psychology, University of Birmingham, Birmingham, B15 2TT, by email: hos.psychology@contacts.bham.ac.uk or by phone on 0121 414 4931

Confidentiality

The confidentiality of participants will be ensured. If published, information on the participant will be presented without reference to their name or any other identifying information. All personal details will be kept separately from the information collected so that it will only be possible to connect results to individuals via a special code. This will ensure that results are kept anonymous. In the unlikely event of any evidence of abuse being identified, this information will be disclosed by the research workers.

Review

The study has been approved by the Ethical Review Committee

Further information

If you would like any more information about the study please contact with [redacted]

Giving consent

Now it is up to you whether you decide that you and your child/the person you care for would like to participate. The decision about whether or not to take part in the study must be 'informed'. This means that anyone making the decision must understand exactly what is involved in the study, what will be required from participants and why.

IMPORTANT:

You need to decide whether your child/the person you care for is able to understand enough about the study to make an 'informed' decision independently about whether or not they would like to participate and to communicate this decision to you. If you are unsure whether or not your child/person you care for is able to understand enough to make a decision independently then we can provide you with some guidelines to help you to assess this A symbol information sheet can also be made available to you if this would be of help.

[redacted]

Please choose from one of the following options:

1. My child/ the person I care for is <u>able to understand</u> what is involved in the study and what will be required from them if they participate and has communicated their decision to me:

If you think that the person is **<u>able</u>** to understand enough about the study in order to make an 'informed' decision and they decide that they would like to participate then please ensure that they complete **Section 1 of <u>Consent Form A</u>** enclosed, or that you complete it with them, on their behalf.

A parent/carer will need to complete **Section 2 of <u>Consent From A</u>** in order to indicate that they also agree to participate in the study. *A symbol information sheet can be made available in order to support your child/person you care for in making this decision if it would be of help.* Please contact the research team if you would like a copy of the symbol consent form or if you need us to adapt this information further, in order to suit your child's needs. Please return the consent form along with the questionnaire pack to us in the prepaid envelope provided.

2. My child/ the person I care for is <u>unable to understand</u> what is involved in the study and what will be required from them if they participate (either because they are too young to understand or because they are unable to understand) and cannot communicate their decision to me:

If you are reading this information on behalf of someone you care for who is under the age of 16 years and you decide that the person ***<u>is not</u>*** able to make an 'informed' and independent decision about whether or not they would like to participate, then we would like to ask you to decide whether or not you think that it is in your child's best interests for them to participate in the study and whether you would like to provide your consent to participation on their behalf. If you would like your child/person you care for to participate in this study, please complete **<u>Consent Form B</u>** enclosed. Please return the consent form along with the questionnaire pack to us in the prepaid envelope provided.

Appendix 2- Consent Forms

UNIVERSITY OF BIRMINGHAM

Consent Form A : For individuals who are able to provide consent to participate in the study

Understanding behaviour and family adjustment in individuals with neurodevelopmental disorders

Study Director: Professor Chris Oliver

SECTION 1: Please complete this section if you are a person with X syndrome:

1. Has somebody else explained the project to you or have you read the information?
   YES/NO
2. Do you understand what the project is about?
   YES/NO
3. Have you asked all of the questions you want?
   YES/NO
4. Have you had your questions answered in a way you understand?
   YES/NO
5. Do you understand it is OK to stop taking part at any time?
   YES/NO
6. Are you happy to take part?
   YES/NO

If any answers are 'no' or you don't want to take part, don't sign your name!

If you do want to take part, you can write your name below

You can also choose if you want to say 'yes' to these questions:

7. If your Dr asks to see your results from this project is that OK?
   YES/NO

8. Are you happy for us to contact you again in the future?
   YES/NO

Your name:_____

Date:_____

The person who explained this project to you needs to sign too. If you are not aged 16 or above, this should be your parent/guardian.

Print name:_____

Sign:_____

Date:_____

SECTION 2: Please complete this section if you are a parent/carer/guardian of a person with X syndrome who has provided their consent to participate in the study.

Please initial box…

1. I confirm that I have read and understood the information sheet dated…. (version….) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. ☐

3. I understand that relevant sections of my child's/person I care for's GP medical notes or records confirming genetic diagnosis and health status may be looked at by members of the Cerebra Centre for Neurodevelopmental Disorders research team at the University of Birmingham, where it is relevant to this research project. I give permission for these individuals to have access to these records. ☐

4. I agree to my child's/person I care for's GP being informed of my participation and that of my child/person I care for's in the study, where access to my child's/person I care for's medical records is required. ☐

5. I agree to take part in the above study. ☐

Optional clause: The statement below is optional:

1. I agree to the University of Birmingham research team sharing my research data with any professionals or clinicians working with me and the person I care for should they request to see them. ☐

Print Name: _____ Telephone number: _____

Address:

_____

Email: ______________________________________

Relationship to participant:________________ Signature: _______________________

Date: __________________

Consent Form B: **For children under the age of 16 who are not able to provide consent.**

Understanding behaviour and family adjustment in individuals with neurodevelopmental disorders

Study Director: Professor Chris Oliver

Please complete this section if you are a parent/ guardian of a child (under 16 years) with X syndrome who is not able to provide consent.

Please initial box…

2. I confirm that I have read and understood the information sheet dated…. (version….) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

3. I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. ☐

4. I understand that relevant sections of my child's/person I care for's GP medical notes or records confirming genetic diagnosis and health status may be looked at by members of the Cerebra Centre for Neurodevelopmental Disorders research team at the University of Birmingham, where it is relevant to this research project. I give permission for these individuals to have access to these records. ☐

5. I agree to my child's/person I care for's GP being informed of my participation and that of my child/person I care for's in the study, where access to my child's/person I care for's medical records is required. ☐

6. I agree to take part in the above study. ☐

Optional clause: The statement below is optional:

6. I agree to the University of Birmingham research team sharing my research data with any professionals or clinicians working with me and the person I care for should they request to see them. ☐

Print Name: _____

Name of person you care for:_____

Address:_____

Email: _____

Telephone number:_____

Relationship to participant: _____

Signature: _____

Date: _____

Appendix 3- Illness Perceptions Questionnaire- Revised for Genetic Syndromes

Parental Understanding and Beliefs

YOUR VIEWS ABOUT YOUR THE PHYSICAL, PSYCHOLOGICAL AND BEHAVIOURAL FEATURES AND SYMPTOMS OF YOUR CHILD'S GENETIC SYNDROME

Listed below are a number of physical, behavioural and psychological features and symptoms that you may or may not have noticed in your child/the person you care for. Please indicate by circling *Yes* or *No*, whether your child/the person you care for, has experienced any of these symptoms or features and whether you believe that these symptoms are related to their genetic syndrome.

	My child/ the person I care for has experienced this symptom/ feature		**This symptom/feature is related to their GS**	
	Yes	**No**	**Yes**	**No**
Difference in physical appearance (facial or other physical differences)				
Learning disability				
Challenging behaviour (e.g. aggression or self-injury, temper outbursts)				
Physical health problems				
Sensory problems (e.g. impaired vision or hearing)				
Neurological problems (e.g. seizures)				
Physical disability				
Autism or autistic-like behaviour				
Mental health problems (e.g. anxiety, depression)				
Sleep problems				
Over or under-eating				
Hyperactivity or hyperactive-like behaviour				

We are interested in your own personal views of how you now see your child's genetic syndrome (GS). Please indicate how much you agree or disagree with the following statements about your child's GS by ticking the appropriate box.

Views about your child/the person you care for…	**Strongly Disagree**	**Disagree**	**Neither agree nor disagree**	**Agree**	**Strongly agree**
Their genetic syndrome is a serious condition					
Their genetic syndrome has major consequences on ***THEIR*** life					
Their genetic syndrome does not have much effect on ***THEIR*** life					
Their GS strongly affects the way others see ***THEM***					
Their genetic syndrome has serious financial consequences					
Their genetic syndrome causes difficulties for those who are close to me					
There is a lot which ***I*** can do to control the symptoms relating to my child's genetic syndrome					
What ***I DO*** can determine whether their symptoms gets better or worse					
The course of my their symptoms and features depends on me					
Nothing I do will affect their genetic syndrome					

I have the power to influence their genetic syndrome					
My actions will have no affect on the outcome of their genetic syndrome					
There is very little that can be done to improve the symptoms/features of their genetic syndrome					
The care I provide will be effective in managing the symptoms/features of their genetic syndrome					
The negative effects of their genetic syndrome can be prevented (avoided) by the care I provide.					
The care I provide can control the symptoms/features of their genetic syndrome.					
There is nothing that can help the symptoms/features of their genetic syndrome.					
The symptoms/features of their genetic syndrome are puzzling to me.					
Their genetic syndrome is a mystery to me.					
I don't understand their genetic syndrome.					
Their genetic syndrome doesn't make any sense to me.					
I have a clear picture or understanding of their genetic syndrome.					
The symptoms of my CHILD'S genetic syndrome change a great deal from day to day.					

The symptoms/features related to their genetic syndrome come and go in cycles.					
Their genetic syndrome is very unpredictable.					
My child goes through cycles in which their symptoms get better and worse.					
I get depressed when I think about their genetic syndrome.					
When I think about their genetic syndrome I get upset.					
Their genetic syndrome makes me feel angry.					
Their genetic syndrome does not worry me.					
Having a child with a genetic syndrome makes me feel anxious.					
Their genetic syndrome makes me feel afraid.					

Appendix 4- Paired Samples T-Test of IPQ-RGS Identity Subscale Items SPSS Output

Paired Samples Statistics

		Mean	N	Std. Deviation	Std. Error Mean
Pair 1	IPQ Identity	8.3091	110	2.29377	.21870
	IPQ_Somatisation	7.9273	110	2.21218	.21092

Paired Samples Correlations

		N	Correlation	Sig.
Pair 1	IPQ Identity & IPQ_Somatisation	110	.731	.000

Paired Samples Test

	Paired Differences					t	df	Sig. (2-tailed)
	Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				
				Lower	Upper			
IPQ Identity - IPQ_Somatisation	.38182	1.65336	.15764	.06938	.69426	2.422	109	.017

Appendix 5- Frequency of IPQ-RGS Identity Scale Item Endorsements SPSS Output

[Difference in physical appearance (facial or other physical differences)] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	97	78.2	78.9	78.9
	No	26	21.0	21.1	100.0
	Total	123	99.2	100.0	
Missing	999	1	.8		
Total		124	100.0		

[Learning disability] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	122	98.4	99.2	99.2
	No	1	.8	.8	100.0
	Total	123	99.2	100.0	
Missing	999	1	.8		
Total		124	100.0		

[Challenging behaviour (e.g. aggression or self-injury, temper outbursts)] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	102	82.3	84.3	84.3
	No	19	15.3	15.7	100.0
	Total	121	97.6	100.0	
Missing	999	3	2.4		
Total		124	100.0		

[Physical health problems] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	98	79.0	80.3	80.3
	No	24	19.4	19.7	100.0
	Total	122	98.4	100.0	
Missing	999	2	1.6		
Total		124	100.0		

[Sensory problems (e.g. impaired vision or hearing)] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	80	64.5	65.6	65.6
	No	42	33.9	34.4	100.0
	Total	122	98.4	100.0	
Missing	999	2	1.6		
Total		124	100.0		

[Neurological problems (e.g. seizures)] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	68	54.8	55.7	55.7
	No	54	43.5	44.3	100.0
	Total	122	98.4	100.0	
Missing	999	2	1.6		
Total		124	100.0		

[Physical disability] [Scale 2]

		Frequency	Percent	Valid Percent	Cumulative Percent
Valid	Yes	102	82.3	83.6	83.6
	No	20	16.1	16.4	100.0
	Total	122	98.4	100.0	
Missing	999	2	1.6		
Total		124	100.0		

Appendix 6- IPQ-RGS Subscale Reliabilities SPSS Output

Scale: IPQ Identity

Case Processing Summary

		N	%
Cases	Valid	110	97.3
	Excluded[a]	3	2.7
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.625	.648	12

Summary Item Statistics

	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	.692	.336	.991	.655	2.946	.030	12

Scale: IPQ Consequence

Case Processing Summary

		N	%
Cases	Valid	113	100.0
	Excluded[a]	0	.0
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.669	.691	6

Summary Item Statistics

	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	4.385	4.080	4.717	.637	1.156	.062	6

Scale Statistics

Mean	Variance	Std. Deviation	N of Items
26.3097	11.001	3.31684	6

Scale: IPQ Personal Control

Case Processing Summary

		N	%
Cases	Valid	113	100.0
	Excluded[a]	0	.0
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.778	.779	5

Summary Item Statistics

	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	2.786	2.460	3.186	.726	1.295	.074	5

Scale Statistics

Mean	Variance	Std. Deviation	N of Items
13.9292	20.834	4.56445	5

Scale: IPQ Treatment Control

Case Processing Summary

		N	%
Cases	Valid	113	100.0
	Excluded[a]	0	.0
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.674	.676	5

Summary Item Statistics

	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	3.350	2.805	3.885	1.080	1.385	.159	5

Scale Statistics

Mean	Variance	Std. Deviation	N of Items
16.7522	12.492	3.53435	5

Scale: IPQ Illness Coherence

Case Processing Summary

		N	%
Cases	Valid	113	100.0
	Excluded[a]	0	.0
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.823	.838	5

Summary Item Statistics

	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	4.030	3.460	4.327	.867	1.251	.114	5

Scale Statistics

Mean	Variance	Std. Deviation	N of Items
20.1504	11.683	3.41797	5

Scale: IPQ Timeline Cyclical

Case Processing Summary

		N	%
Cases	Valid	113	100.0
	Excluded[a]	0	.0
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.820	.820	4

Summary Item Statistics

	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	2.845	2.531	3.195	.664	1.262	.075	4

Scale Statistics

Mean	Variance	Std. Deviation	N of Items
11.38	13.434	3.665	4

Scale: IPQ Emotional Representation

Case Processing Summary

		N	%
Cases	Valid	113	100.0
	Excluded[a]	0	.0
	Total	113	100.0

a. Listwise deletion based on all variables in the procedure.

Reliability Statistics

Cronbach's Alpha	Cronbach's Alpha Based on Standardized Items	N of Items
.849	.851	6

Summary Item Statistics

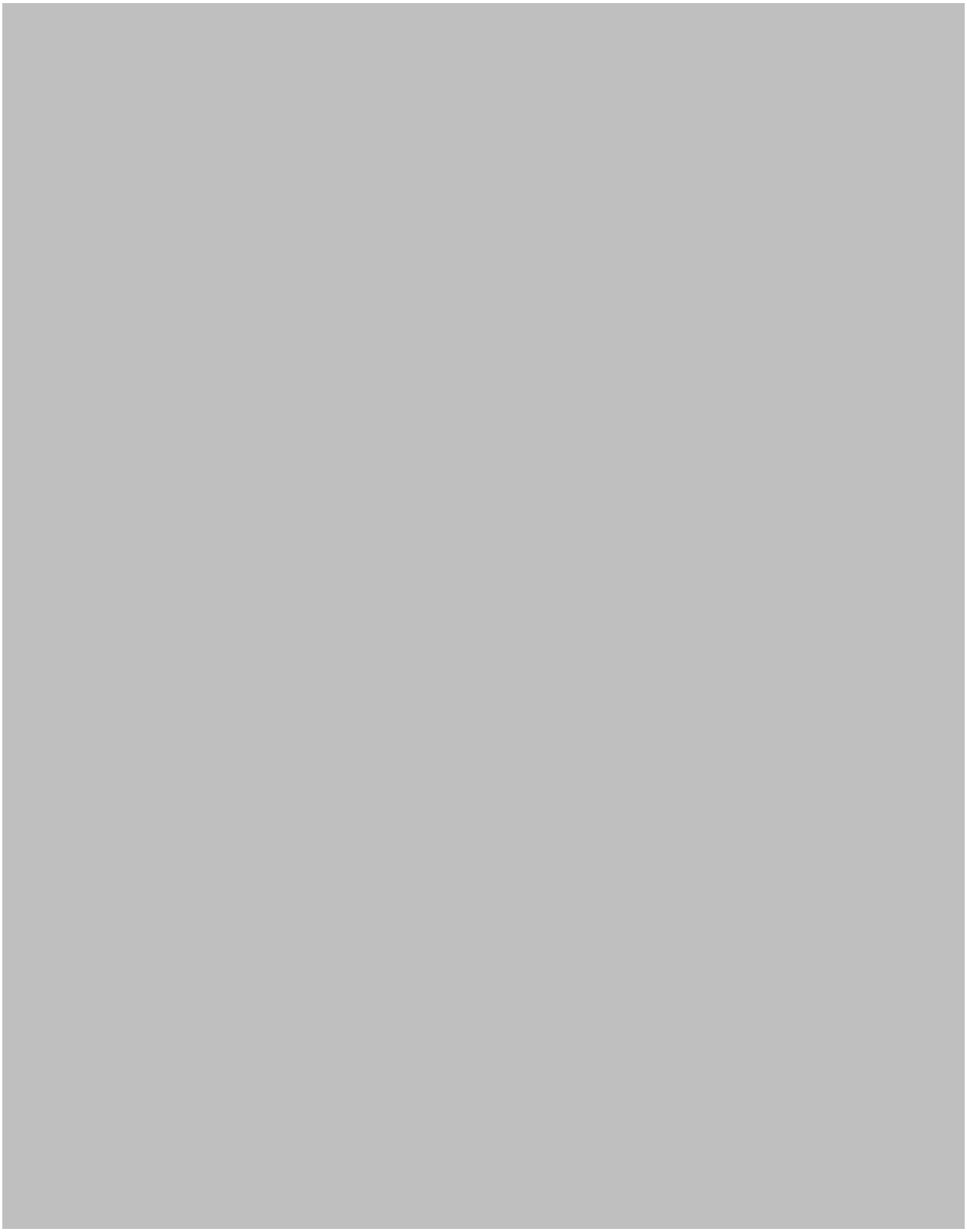
	Mean	Minimum	Maximum	Range	Maximum / Minimum	Variance	N of Items
Item Means	3.263	2.681	4.018	1.336	1.498	.209	6

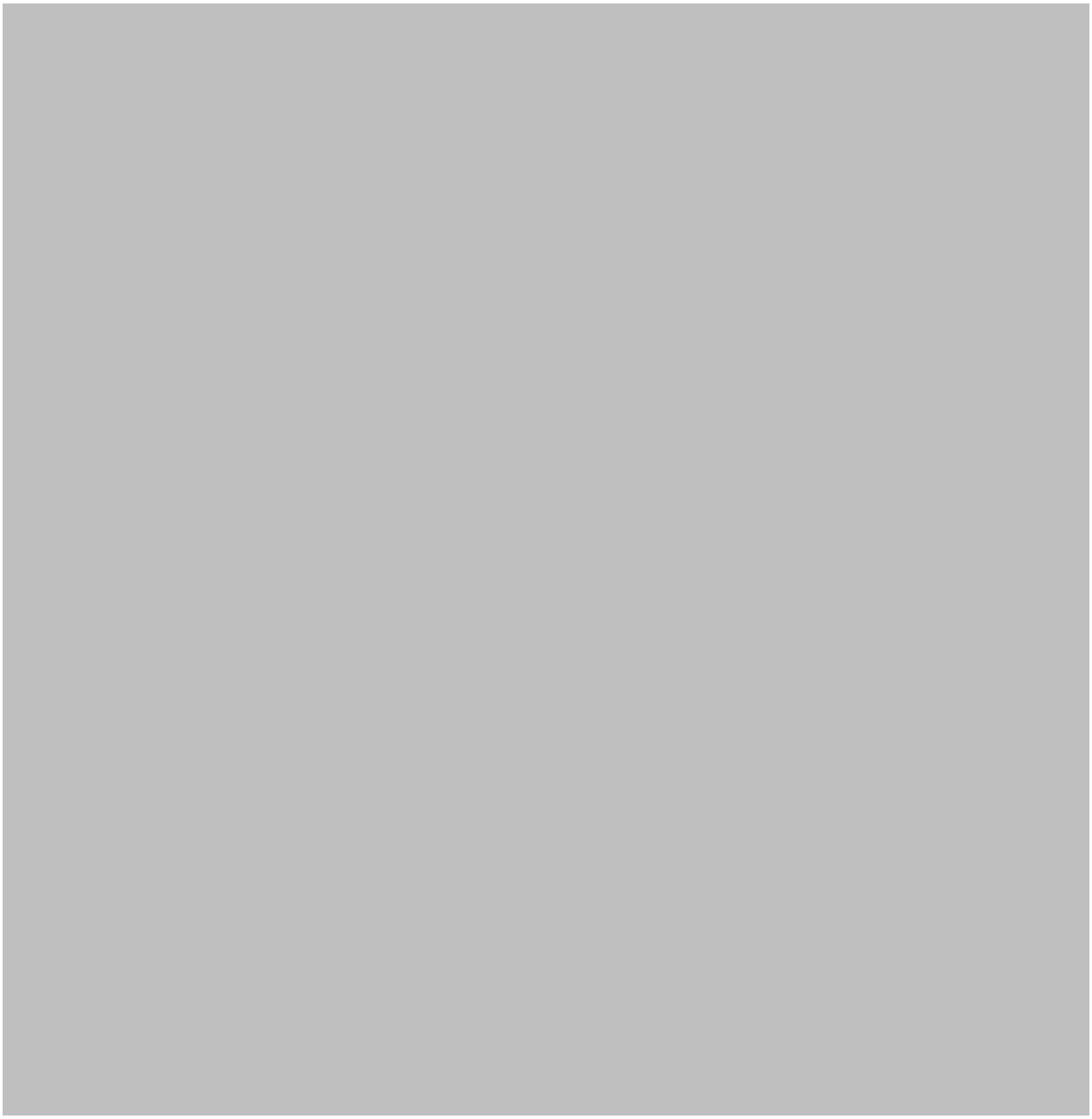
Scale Statistics

Mean	Variance	Std. Deviation	N of Items
19.5752	27.657	5.25901	6

Appendix 7- Brief COPE (Carver, 1997).

Brief COPE





Appendix 8- Hospital Anxiety and Depression Scale (*HADS*: Zigmond and Snaith, 1983)©

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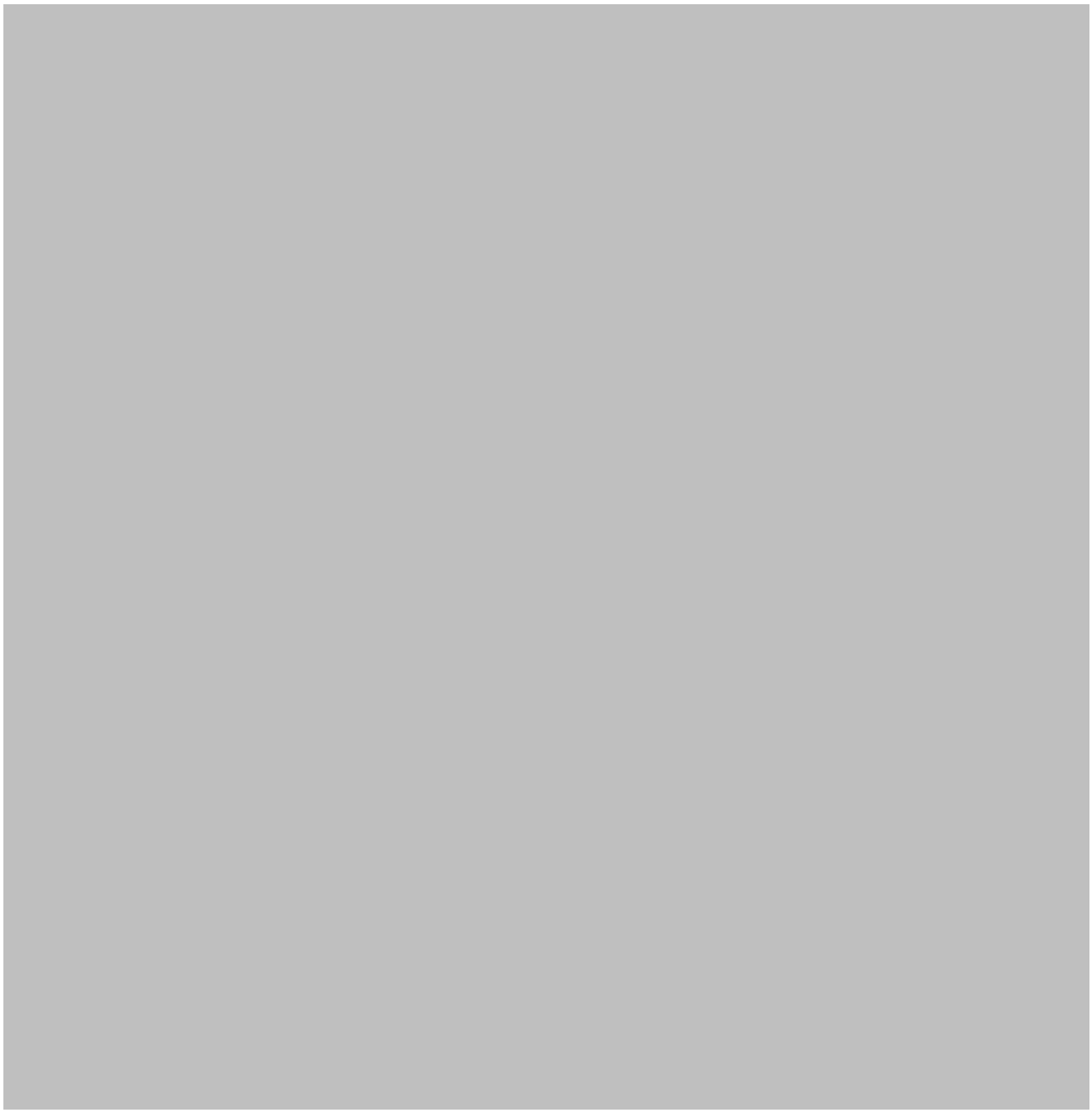
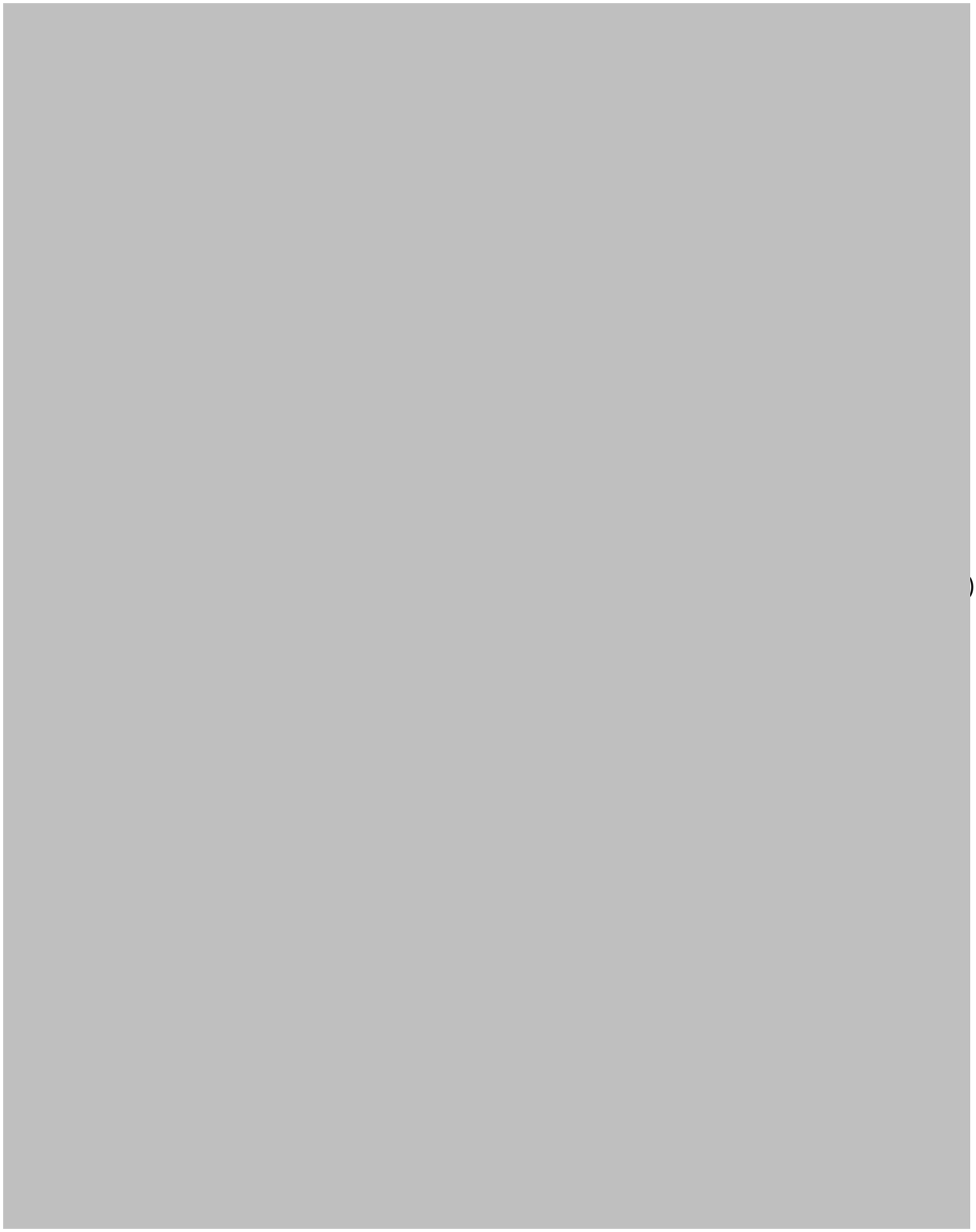
Appendix 9- The Positive and Negative Affect Scale (PANAS; Watson et al., 1988).

Positive and Negative Affect Schedule



Appendix 10- The Wessex (Kushlick et al., 1973).

WESSEX Questionnaire



Appendix 11- Univariate Associations with Anxiety and Depression Correlations Matrix SPSS Output

Descriptive Statistics

	Mean	Std. Deviation	N
HADS Total Anxiety Score	9.58	4.661	113
HADS Total Depression Score	6.50	4.007	113
PANAS Positive Affect	30.92	9.247	113
PANAS Negative Affect	20.30	8.546	113
IPQ Identity Scale Total Score	15.6909	2.29377	110
IPQ Consequence	26.3097	3.31684	113
IPQ Personal Control (Item 12 removed)	13.9292	4.56445	113
IPQ Treatment Control	16.7522	3.53435	113
IPQ Illness Coherence	20.1504	3.41797	113
IPQ Timeline Cyclical	11.3805	3.66528	113
IPQ Emotional Representation	19.5752	5.25901	113
COPE Self Distraction	3.72	1.454	113
COPE Active Coping	5.76	1.829	113
COPE Denial	2.21	.661	113
COPE Substance Use	2.81	1.455	113
COPE Emotional Support	4.53	1.727	113
COPE Instrumental Support	5.12	1.657	113
COPE Behavioural Disengagement	2.38	.783	113
COPE Venting	3.50	1.324	113
COPE Positive Reframing	5.28	1.740	113
COPE Planning	5.82	1.784	113
COPE Humour	3.64	1.717	113
COPE Acceptance	6.91	1.430	113
COPE Religion	2.96	1.658	113
COPE Self Blame	3.53	1.637	113
age	9.9646	4.08597	113
Wessex self help total score (wsxg+wsxh+wsxi)	135.2655	289.80474	113

		HADS Total Anxiety Score	HADS Total Depression Score	PANAS Positive Affect	PANAS Negative Affect	IPQ Identity Scale Total Score	IPQ Consequence	IPQ Personal Control (Item 12 removed)
HADS Total Anxiety Score	Pearson Correlation	1	.715**	-.399**	.710**	-.295**	.188*	-.091
	Sig. (2-tailed)		.000	.000	.000	.002	.047	.339
	N	113	113	113	113	110	113	113
HADS Total Depression Score	Pearson Correlation	.715**	1	-.517**	.586**	-.233*	.152	-.209*
	Sig. (2-tailed)	.000		.000	.000	.014	.107	.026
	N	113	113	113	113	110	113	113
PANAS Positive Affect	Pearson Correlation	-.399**	-.517**	1	-.318**	.035	-.040	.164
	Sig. (2-tailed)	.000	.000		.001	.718	.677	.082
	N	113	113	113	113	110	113	113
PANAS Negative Affect	Pearson Correlation	.710**	.586**	-.318**	1	-.200*	.177	-.132
	Sig. (2-tailed)	.000	.000	.001		.036	.061	.163
	N	113	113	113	113	110	113	113
IPQ Identity Scale Total Score	Pearson Correlation	-.295**	-.233*	.035	-.200*	1	-.367**	.018
	Sig. (2-tailed)	.002	.014	.718	.036		.000	.854
	N	110	110	110	110	110	110	110
IPQ Consequence	Pearson Correlation	.188*	.152	-.040	.177	-.367**	1	-.210*
	Sig. (2-tailed)	.047	.107	.677	.061	.000		.026
	N	113	113	113	113	110	113	113
IPQ Personal Control (Item	Pearson Correlation	-.091	-.209*	.164	-.132	.018	-.210*	1

12 removed)	Sig. (2-tailed)	.339	.026	.082	.163	.854	.026	
	N	113	113	113	113	110	113	113
IPQ Treatment Control	Pearson Correlation	-.045	-.042	.149	-.087	-.065	-.162	.563**
	Sig. (2-tailed)	.637	.661	.115	.361	.499	.087	.000
	N	113	113	113	113	110	113	113
IPQ Illness Coherence	Pearson Correlation	-.078	-.252**	.239*	-.090	-.160	.212*	.014
	Sig. (2-tailed)	.410	.007	.011	.345	.096	.024	.884
	N	113	113	113	113	110	113	113
IPQ Timeline Cyclical	Pearson Correlation	.263**	.228*	.017	.169	-.236*	.097	.035
	Sig. (2-tailed)	.005	.015	.856	.073	.013	.305	.711
	N	113	113	113	113	110	113	113
IPQ Emotional Representation	Pearson Correlation	.526**	.410**	-.194*	.517**	-.062	.271**	-.107
	Sig. (2-tailed)	.000	.000	.040	.000	.519	.004	.258
	N	113	113	113	113	110	113	113
COPE Self Distraction	Pearson Correlation	.137	.127	.075	.255**	-.185	.207*	-.138
	Sig. (2-tailed)	.146	.180	.432	.006	.053	.028	.146
	N	113	113	113	113	110	113	113
COPE Active Coping	Pearson Correlation	.143	-.003	.393**	.057	-.260**	.137	.079
	Sig. (2-tailed)	.131	.973	.000	.551	.006	.147	.404
	N	113	113	113	113	110	113	113
COPE Denial	Pearson Correlation	.093	.196*	-.135	.265**	.164	.023	-.030
	Sig. (2-	.326	.038	.155	.005	.087	.812	.748

	tailed)							
	N	113	113	113	113	110	113	113
COPE Substance Use	Pearson Correlation	.308**	.330**	-.312**	.407**	-.208*	.112	-.188*
	Sig. (2-tailed)	.001	.000	.001	.000	.029	.238	.047
	N	113	113	113	113	110	113	113
COPE Emotional Support	Pearson Correlation	-.059	-.255**	.253**	-.020	-.077	.083	.047
	Sig. (2-tailed)	.532	.006	.007	.834	.427	.381	.623
	N	113	113	113	113	110	113	113
COPE Instrumental Support	Pearson Correlation	.138	.032	.227*	.114	-.206*	.146	.053
	Sig. (2-tailed)	.144	.739	.016	.231	.031	.122	.577
	N	113	113	113	113	110	113	113
COPE Behavioural Disengagement	Pearson Correlation	.333**	.363**	-.352**	.489**	-.106	.106	-.272**
	Sig. (2-tailed)	.000	.000	.000	.000	.268	.266	.004
	N	113	113	113	113	110	113	113
COPE Venting	Pearson Correlation	.205*	.169	.080	.331**	-.339**	.201*	-.167
	Sig. (2-tailed)	.029	.074	.401	.000	.000	.033	.077
	N	113	113	113	113	110	113	113
COPE Positive Reframing	Pearson Correlation	.015	-.042	.405**	-.034	-.018	.059	.188*
	Sig. (2-tailed)	.875	.658	.000	.721	.848	.535	.046
	N	113	113	113	113	110	113	113
COPE Planning	Pearson Correlation	.191*	.121	.244**	.202*	-.351**	.234*	-.032
	Sig. (2-tailed)	.043	.202	.009	.032	.000	.013	.735

	N	113	113	113	113	110	113	113
COPE Humour	Pearson Correlation	-.002	-.045	.239*	.073	-.027	.014	.066
	Sig. (2-tailed)	.987	.636	.011	.441	.783	.886	.486
	N	113	113	113	113	110	113	113
COPE Acceptance	Pearson Correlation	.056	-.039	.318**	.036	-.176	.132	-.015
	Sig. (2-tailed)	.556	.682	.001	.707	.065	.164	.878
	N	113	113	113	113	110	113	113
COPE Religion	Pearson Correlation	-.060	-.105	.189*	-.018	.040	.025	.008
	Sig. (2-tailed)	.530	.269	.046	.849	.679	.795	.934
	N	113	113	113	113	110	113	113
COPE Self Blame	Pearson Correlation	.278**	.390**	-.141	.461**	-.165	.254**	-.094
	Sig. (2-tailed)	.003	.000	.138	.000	.085	.007	.321
	N	113	113	113	113	110	113	113
age	Pearson Correlation	-.149	.019	.018	-.133	-.138	.017	-.210*
	Sig. (2-tailed)	.114	.846	.853	.159	.152	.861	.025
	N	113	113	113	113	110	113	113
Wessex self help total score (wsxg+wsxh+wsxi)	Pearson Correlation	-.101	-.192*	.132	-.094	-.160	.051	.149
	Sig. (2-tailed)	.285	.042	.163	.324	.096	.594	.116
	N	113	113	113	113	110	113	113

		IPQ Treatment Control	IPQ Illness Coherence	IPQ Timeline Cyclical	IPQ Emotional Representation	COPE Self Distraction	COPE Active Coping	COPE Denial
HADS Total Anxiety Score	Pearson Correlation	-.045	-.078	.263**	.526**	.137	.143	.093
	Sig. (2-tailed)	.637	.410	.005	.000	.146	.131	.326
	N	113	113	113	113	113	113	113
HADS Total Depression Score	Pearson Correlation	-.042	-.252**	.228*	.410**	.127	-.003	.196*
	Sig. (2-tailed)	.661	.007	.015	.000	.180	.973	.038
	N	113	113	113	113	113	113	113
PANAS Positive Affect	Pearson Correlation	.149	.239*	.017	-.194*	.075	.393**	-.135
	Sig. (2-tailed)	.115	.011	.856	.040	.432	.000	.155
	N	113	113	113	113	113	113	113
PANAS Negative Affect	Pearson Correlation	-.087	-.090	.169	.517**	.255**	.057	.265**
	Sig. (2-tailed)	.361	.345	.073	.000	.006	.551	.005
	N	113	113	113	113	113	113	113
IPQ Identity Scale Total Score	Pearson Correlation	-.065	-.160	-.236*	-.062	-.185	-.260**	.164
	Sig. (2-tailed)	.499	.096	.013	.519	.053	.006	.087
	N	110	110	110	110	110	110	110
IPQ Consequence	Pearson Correlation	-.162	.212*	.097	.271**	.207*	.137	.023
	Sig. (2-tailed)	.087	.024	.305	.004	.028	.147	.812
	N	113	113	113	113	113	113	113

IPQ Personal Control (Item 12 removed)	Pearson Correlation	.563**	.014	.035	-.107	-.138	.079	-.030
	Sig. (2-tailed)	.000	.884	.711	.258	.146	.404	.748
	N	113	113	113	113	113	113	113
IPQ Treatment Control	Pearson Correlation	1	.033	.105	-.051	-.087	.155	-.084
	Sig. (2-tailed)		.731	.267	.593	.361	.101	.375
	N	113	113	113	113	113	113	113
IPQ Illness Coherence	Pearson Correlation	.033	1	-.099	-.250**	.098	.102	-.319**
	Sig. (2-tailed)	.731		.295	.008	.300	.285	.001
	N	113	113	113	113	113	113	113
IPQ Timeline Cyclical	Pearson Correlation	.105	-.099	1	.152	.101	.022	.125
	Sig. (2-tailed)	.267	.295		.109	.288	.820	.188
	N	113	113	113	113	113	113	113
IPQ Emotional Representation	Pearson Correlation	-.051	-.250**	.152	1	.215*	.102	.275**
	Sig. (2-tailed)	.593	.008	.109		.022	.284	.003
	N	113	113	113	113	113	113	113
COPE Self Distraction	Pearson Correlation	-.087	.098	.101	.215*	1	.276**	.202*
	Sig. (2-tailed)	.361	.300	.288	.022		.003	.031
	N	113	113	113	113	113	113	113
COPE Active Coping	Pearson Correlation	.155	.102	.022	.102	.276**	1	-.105
	Sig. (2-tailed)	.101	.285	.820	.284	.003		.267
	N	113	113	113	113	113	113	113

COPE Denial	Pearson Correlation	-.084	-.319**	.125	.275**	.202*	-.105	1
	Sig. (2-tailed)	.375	.001	.188	.003	.031	.267	
	N	113	113	113	113	113	113	113
COPE Substance Use	Pearson Correlation	-.054	.060	.085	.253**	.199*	.087	.088
	Sig. (2-tailed)	.569	.531	.369	.007	.035	.358	.355
	N	113	113	113	113	113	113	113
COPE Emotional Support	Pearson Correlation	.080	.237*	.041	.033	.334**	.309**	.002
	Sig. (2-tailed)	.398	.011	.665	.729	.000	.001	.983
	N	113	113	113	113	113	113	113
COPE Instrumental Support	Pearson Correlation	.165	.218*	.090	.133	.295**	.428**	-.006
	Sig. (2-tailed)	.081	.021	.344	.161	.002	.000	.948
	N	113	113	113	113	113	113	113
COPE Behavioural Disengagement	Pearson Correlation	-.288**	-.048	-.048	.300**	.339**	-.061	.377**
	Sig. (2-tailed)	.002	.612	.615	.001	.000	.523	.000
	N	113	113	113	113	113	113	113
COPE Venting	Pearson Correlation	-.035	.082	.139	.288**	.704**	.319**	.113
	Sig. (2-tailed)	.716	.388	.141	.002	.000	.001	.232
	N	113	113	113	113	113	113	113
COPE Positive Reframing	Pearson Correlation	.113	.083	.095	-.110	.247**	.403**	.040
	Sig. (2-tailed)	.233	.383	.317	.247	.008	.000	.671
	N	113	113	113	113	113	113	113

COPE Planning	Pearson Correlation	.091	.081	-.014	.156	.418**	.562**	-.036
	Sig. (2-tailed)	.339	.396	.881	.100	.000	.000	.705
	N	113	113	113	113	113	113	113
COPE Humour	Pearson Correlation	-.006	.011	.012	-.007	.302**	.237*	-.002
	Sig. (2-tailed)	.949	.909	.898	.939	.001	.012	.981
	N	113	113	113	113	113	113	113
COPE Acceptance	Pearson Correlation	-.070	.153	.063	.086	.147	.347**	.001
	Sig. (2-tailed)	.463	.107	.510	.363	.121	.000	.990
	N	113	113	113	113	113	113	113
COPE Religion	Pearson Correlation	.076	-.021	.171	.067	.000	.083	.048
	Sig. (2-tailed)	.422	.824	.070	.482	.996	.384	.616
	N	113	113	113	113	113	113	113
COPE Self Blame	Pearson Correlation	-.062	-.003	.145	.414**	.165	-.056	.192*
	Sig. (2-tailed)	.515	.973	.127	.000	.081	.558	.042
	N	113	113	113	113	113	113	113
age	Pearson Correlation	-.116	-.080	-.077	-.227*	.024	-.058	-.129
	Sig. (2-tailed)	.220	.399	.416	.016	.802	.538	.172
	N	113	113	113	113	113	113	113
Wessex self help total score (wsxg+wsxh+wsxi)	Pearson Correlation	.032	.223*	.011	-.239*	-.026	.163	-.072
	Sig. (2-tailed)	.736	.017	.908	.011	.788	.084	.448
	N	113	113	113	113	113	113	113

		COPE Substance Use	COPE Emotional Support	COPE Instrumental Support	COPE Behavioural Disengagement	COPE Venting	COPE Positive Reframing	COPE Planning
HADS Total Anxiety Score	Pearson Correlation	.308**	-.059	.138	.333**	.205*	.015	.191*
	Sig. (2-tailed)	.001	.532	.144	.000	.029	.875	.043
	N	113	113	113	113	113	113	113
HADS Total Depression Score	Pearson Correlation	.330**	-.255**	.032	.363**	.169	-.042	.121
	Sig. (2-tailed)	.000	.006	.739	.000	.074	.658	.202
	N	113	113	113	113	113	113	113
PANAS Positive Affect	Pearson Correlation	-.312**	.253**	.227*	-.352**	.080	.405**	.244**
	Sig. (2-tailed)	.001	.007	.016	.000	.401	.000	.009
	N	113	113	113	113	113	113	113
PANAS Negative Affect	Pearson Correlation	.407**	-.020	.114	.489**	.331**	-.034	.202*
	Sig. (2-tailed)	.000	.834	.231	.000	.000	.721	.032
	N	113	113	113	113	113	113	113
IPQ Identity Scale Total Score	Pearson Correlation	-.208*	-.077	-.206*	-.106	-.339**	-.018	-.351**
	Sig. (2-tailed)	.029	.427	.031	.268	.000	.848	.000
	N	110	110	110	110	110	110	110
IPQ Consequence	Pearson Correlation	.112	.083	.146	.106	.201*	.059	.234*
	Sig. (2-tailed)	.238	.381	.122	.266	.033	.535	.013
	N	113	113	113	113	113	113	113
IPQ Personal Control (Item 12 removed)	Pearson Correlation	-.188*	.047	.053	-.272**	-.167	.188*	-.032

	Sig. (2-tailed)	.047	.623	.577	.004	.077	.046	.735
	N	113	113	113	113	113	113	113
IPQ Treatment Control	Pearson Correlation	-.054	.080	.165	-.288**	-.035	.113	.091
	Sig. (2-tailed)	.569	.398	.081	.002	.716	.233	.339
	N	113	113	113	113	113	113	113
IPQ Illness Coherence	Pearson Correlation	.060	.237*	.218*	-.048	.082	.083	.081
	Sig. (2-tailed)	.531	.011	.021	.612	.388	.383	.396
	N	113	113	113	113	113	113	113
IPQ Timeline Cyclical	Pearson Correlation	.085	.041	.090	-.048	.139	.095	-.014
	Sig. (2-tailed)	.369	.665	.344	.615	.141	.317	.881
	N	113	113	113	113	113	113	113
IPQ Emotional Representation	Pearson Correlation	.253**	.033	.133	.300**	.288**	-.110	.156
	Sig. (2-tailed)	.007	.729	.161	.001	.002	.247	.100
	N	113	113	113	113	113	113	113
COPE Self Distraction	Pearson Correlation	.199*	.334**	.295**	.339**	.704**	.247**	.418**
	Sig. (2-tailed)	.035	.000	.002	.000	.000	.008	.000
	N	113	113	113	113	113	113	113
COPE Active Coping	Pearson Correlation	.087	.309**	.428**	-.061	.319**	.403**	.562**
	Sig. (2-tailed)	.358	.001	.000	.523	.001	.000	.000
	N	113	113	113	113	113	113	113
COPE Denial	Pearson Correlation	.088	.002	-.006	.377**	.113	.040	-.036
	Sig. (2-	.355	.983	.948	.000	.232	.671	.705

	tailed)							
	N	113	113	113	113	113	113	113
COPE Substance Use	Pearson Correlation	1	-.014	.009	.408**	.248**	-.088	.149
	Sig. (2-tailed)		.886	.925	.000	.008	.352	.116
	N	113	113	113	113	113	113	113
COPE Emotional Support	Pearson Correlation	-.014	1	.577**	-.078	.395**	.264**	.439**
	Sig. (2-tailed)	.886		.000	.411	.000	.005	.000
	N	113	113	113	113	113	113	113
COPE Instrumental Support	Pearson Correlation	.009	.577**	1	-.096	.430**	.178	.511**
	Sig. (2-tailed)	.925	.000		.312	.000	.060	.000
	N	113	113	113	113	113	113	113
COPE Behavioural Disengagement	Pearson Correlation	.408**	-.078	-.096	1	.325**	-.119	-.028
	Sig. (2-tailed)	.000	.411	.312		.000	.209	.768
	N	113	113	113	113	113	113	113
COPE Venting	Pearson Correlation	.248**	.395**	.430**	.325**	1	.090	.461**
	Sig. (2-tailed)	.008	.000	.000	.000		.345	.000
	N	113	113	113	113	113	113	113
COPE Positive Reframing	Pearson Correlation	-.088	.264**	.178	-.119	.090	1	.249**
	Sig. (2-tailed)	.352	.005	.060	.209	.345		.008
	N	113	113	113	113	113	113	113
COPE Planning	Pearson Correlation	.149	.439**	.511**	-.028	.461**	.249**	1
	Sig. (2-tailed)	.116	.000	.000	.768	.000	.008	

	N	113	113	113	113	113	113	113
COPE Humour	Pearson Correlation	.051	.279**	.172	-.023	.217*	.241*	.221*
	Sig. (2-tailed)	.589	.003	.069	.812	.021	.010	.019
	N	113	113	113	113	113	113	113
COPE Acceptance	Pearson Correlation	.035	.229*	.272**	-.049	.236*	.362**	.442**
	Sig. (2-tailed)	.713	.015	.004	.603	.012	.000	.000
	N	113	113	113	113	113	113	113
COPE Religion	Pearson Correlation	-.029	.119	.177	.059	.069	.155	-.023
	Sig. (2-tailed)	.763	.210	.061	.537	.467	.101	.807
	N	113	113	113	113	113	113	113
COPE Self Blame	Pearson Correlation	.173	.029	.092	.329**	.199*	.000	.121
	Sig. (2-tailed)	.067	.762	.330	.000	.035	1.000	.201
	N	113	113	113	113	113	113	113
age	Pearson Correlation	.155	-.076	-.098	.041	.025	-.015	-.035
	Sig. (2-tailed)	.101	.425	.300	.670	.795	.875	.712
	N	113	113	113	113	113	113	113
Wessex self help total score (wsxg+wsxh+wsxi)	Pearson Correlation	-.137	-.001	-.060	-.188*	.011	.063	.099
	Sig. (2-tailed)	.147	.995	.530	.046	.908	.505	.297
	N	113	113	113	113	113	113	113

		COPE Humour	COPE Acceptance	COPE Religion	COPE Self Blame	age	Wessex self help total score (wsxg+wsxh+wsxi)
HADS Total Anxiety Score	Pearson Correlation	-.002	.056	-.060	.278**	-.149	-.101
	Sig. (2-tailed)	.987	.556	.530	.003	.114	.285
	N	113	113	113	113	113	113
HADS Total Depression Score	Pearson Correlation	-.045	-.039	-.105	.390**	.019	-.192*
	Sig. (2-tailed)	.636	.682	.269	.000	.846	.042
	N	113	113	113	113	113	113
PANAS Positive Affect	Pearson Correlation	.239*	.318**	.189*	-.141	.018	.132
	Sig. (2-tailed)	.011	.001	.046	.138	.853	.163
	N	113	113	113	113	113	113
PANAS Negative Affect	Pearson Correlation	.073	.036	-.018	.461**	-.133	-.094
	Sig. (2-tailed)	.441	.707	.849	.000	.159	.324
	N	113	113	113	113	113	113
IPQ Identity Scale Total Score	Pearson Correlation	-.027	-.176	.040	-.165	-.138	-.160
	Sig. (2-tailed)	.783	.065	.679	.085	.152	.096
	N	110	110	110	110	110	110
IPQ Consequence	Pearson Correlation	.014	.132	.025	.254**	.017	.051
	Sig. (2-tailed)	.886	.164	.795	.007	.861	.594
	N	113	113	113	113	113	113
IPQ Personal Control (Item 12 removed)	Pearson Correlation	.066	-.015	.008	-.094	-.210*	.149
	Sig. (2-tailed)	.486	.878	.934	.321	.025	.116

	N	113	113	113	113	113	113
IPQ Treatment Control	Pearson Correlation	-.006	-.070	.076	-.062	-.116	.032
	Sig. (2-tailed)	.949	.463	.422	.515	.220	.736
	N	113	113	113	113	113	113
IPQ Illness Coherence	Pearson Correlation	.011	.153	-.021	-.003	-.080	.223*
	Sig. (2-tailed)	.909	.107	.824	.973	.399	.017
	N	113	113	113	113	113	113
IPQ Timeline Cyclical	Pearson Correlation	.012	.063	.171	.145	-.077	.011
	Sig. (2-tailed)	.898	.510	.070	.127	.416	.908
	N	113	113	113	113	113	113
IPQ Emotional Representation	Pearson Correlation	-.007	.086	.067	.414**	-.227*	-.239*
	Sig. (2-tailed)	.939	.363	.482	.000	.016	.011
	N	113	113	113	113	113	113
COPE Self Distraction	Pearson Correlation	.302**	.147	.000	.165	.024	-.026
	Sig. (2-tailed)	.001	.121	.996	.081	.802	.788
	N	113	113	113	113	113	113
COPE Active Coping	Pearson Correlation	.237*	.347**	.083	-.056	-.058	.163
	Sig. (2-tailed)	.012	.000	.384	.558	.538	.084
	N	113	113	113	113	113	113
COPE Denial	Pearson Correlation	-.002	.001	.048	.192*	-.129	-.072
	Sig. (2-tailed)	.981	.990	.616	.042	.172	.448
	N	113	113	113	113	113	113
COPE Substance Use	Pearson Correlation	.051	.035	-.029	.173	.155	-.137

	Sig. (2-tailed)	.589	.713	.763	.067	.101	.147
	N	113	113	113	113	113	113
COPE Emotional Support	Pearson Correlation	.279**	.229*	.119	.029	-.076	-.001
	Sig. (2-tailed)	.003	.015	.210	.762	.425	.995
	N	113	113	113	113	113	113
COPE Instrumental Support	Pearson Correlation	.172	.272**	.177	.092	-.098	-.060
	Sig. (2-tailed)	.069	.004	.061	.330	.300	.530
	N	113	113	113	113	113	113
COPE Behavioural Disengagement	Pearson Correlation	-.023	-.049	.059	.329**	.041	-.188*
	Sig. (2-tailed)	.812	.603	.537	.000	.670	.046
	N	113	113	113	113	113	113
COPE Venting	Pearson Correlation	.217*	.236*	.069	.199*	.025	.011
	Sig. (2-tailed)	.021	.012	.467	.035	.795	.908
	N	113	113	113	113	113	113
COPE Positive Reframing	Pearson Correlation	.241*	.362**	.155	.000	-.015	.063
	Sig. (2-tailed)	.010	.000	.101	1.000	.875	.505
	N	113	113	113	113	113	113
COPE Planning	Pearson Correlation	.221*	.442**	-.023	.121	-.035	.099
	Sig. (2-tailed)	.019	.000	.807	.201	.712	.297
	N	113	113	113	113	113	113
COPE Humour	Pearson Correlation	1	.256**	-.064	.028	.024	-.070
	Sig. (2-tailed)		.006	.500	.770	.804	.459
	N	113	113	113	113	113	113

COPE Acceptance	Pearson Correlation	.256**	1	.059	.058	.045	.128
	Sig. (2-tailed)	.006		.535	.539	.634	.176
	N	113	113	113	113	113	113
COPE Religion	Pearson Correlation	-.064	.059	1	.030	.054	-.034
	Sig. (2-tailed)	.500	.535		.752	.571	.724
	N	113	113	113	113	113	113
COPE Self Blame	Pearson Correlation	.028	.058	.030	1	-.081	-.045
	Sig. (2-tailed)	.770	.539	.752		.392	.636
	N	113	113	113	113	113	113
age	Pearson Correlation	.024	.045	.054	-.081	1	-.169
	Sig. (2-tailed)	.804	.634	.571	.392		.074
	N	113	113	113	113	113	113
Wessex self help total score (wsxg+wsxh+wsxi)	Pearson Correlation	-.070	.128	-.034	-.045	-.169	1
	Sig. (2-tailed)	.459	.176	.724	.636	.074	
	N	113	113	113	113	113	113

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

*. Correlation is significant at the 0.05 level (2-tailed).

Appendix 12- Logistic Regression Analysis with Anxiety as Outcome SPSS Output

Case Processing Summary

Unweighted Cases[a]		N	Percent
Selected Cases	Included in Analysis	110	97.3
	Missing Cases	3	2.7
	Total	113	100.0
Unselected Cases		0	.0
Total		113	100.0

a. If weight is in effect, see classification table for the total number of cases.

Dependent Variable Encoding

Original Value	Internal Value
Normal	0
Above Clinical	1

Block 0: Beginning Block

Iteration History[a,b,c]

Iteration		-2 Log likelihood	Coefficients
			Constant
Step 0	1	140.506	.655
	2	140.491	.679
	3	140.491	.680

a. Constant is included in the model.

b. Initial -2 Log Likelihood: 140.491

c. Estimation terminated at iteration number 3 because parameter estimates changed by less than .001.

Classification Table[a,b]

	Observed		Predicted		
			HADS Anxiety Split into groups of Normal and Above Clinical		Percentage Correct
			Normal	Above Clinical	
Step 0	HADS Anxiety Split into groups of Normal and Above Clinical	Normal	0	37	.0
		Above Clinical	0	73	100.0
	Overall Percentage				66.4

a. Constant is included in the model.

b. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 0	Constant	.680	.202	11.339	1	.001	1.973

Variables not in the Equation

			Score	df	Sig.
Step 0	Variables	IPQ_Identity	6.317	1	.012
		IPQ_TimelineCyclical	9.155	1	.002
		IPQ_EmotionalRep	21.123	1	.000
		COPE_SU	11.060	1	.001
		COPE_BD	8.161	1	.004
		COPE_SB	4.327	1	.038
	Overall Statistics		33.722	6	.000

Block 1: Method = Enter

Iteration History[a,b,c,d]

Iteration		-2 Log likelihood	Coefficients						
			Constant	IPQ Identity	IPQ Timeline Cyclical	IPQ Emotional Rep	COPE_SU	COPE_BD	COPE_SB
Step 1	1	105.397	-4.922	.115	.103	.125	.183	.306	-.070
	2	99.060	-7.222	.152	.132	.161	.413	.607	-.092
	3	97.598	-8.622	.166	.142	.169	.641	.863	-.097
	4	97.498	-9.073	.170	.145	.172	.723	.944	-.099
	5	97.497	-9.107	.170	.146	.172	.730	.950	-.100
	6	97.497	-9.107	.170	.146	.172	.730	.950	-.100

a. Method: Enter

b. Constant is included in the model.

c. Initial -2 Log Likelihood: 140.491

d. Estimation terminated at iteration number 6 because parameter estimates changed by less than .001.

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	42.993	6	.000
	Block	42.993	6	.000
	Model	42.993	6	.000

Model Summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	97.497[a]	.324	.449

a. Estimation terminated at iteration number 6 because parameter estimates changed by less than .001.

Hosmer and Lemeshow Test

Step	Chi-square	df	Sig.
1	5.078	8	.749

Contingency Table for Hosmer and Lemeshow Test

		HADS Anxiety Split into groups of Normal and Above Clinical = Normal		HADS Anxiety Split into groups of Normal and Above Clinical = Above Clinical		Total
		Observed	Expected	Observed	Expected	
Step 1	1	10	9.253	1	1.747	11
	2	8	7.579	3	3.421	11
	3	4	6.053	7	4.947	11
	4	6	4.971	5	6.029	11
	5	4	3.918	7	7.082	11
	6	1	2.566	10	8.434	11
	7	2	1.365	9	9.635	11
	8	1	.839	10	10.161	11
	9	1	.388	10	10.612	11
	10	0	.068	11	10.932	11

Classification Table[a]

Observed			Predicted		
			HADS Anxiety Split into groups of Normal and Above Clinical		Percentage Correct
			Normal	Above Clinical	
Step 1	HADS Anxiety Split into groups of Normal and Above Clinical	Normal	22	15	59.5
		Above Clinical	10	63	86.3
	Overall Percentage				77.3

a. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I.for EXP(B)	
								Lower	Upper
Step 1[a]	IPQ_Identity	.170	.124	1.877	1	.171	1.186	.929	1.513
	IPQ_TimelineCyclical	.146	.075	3.771	1	.052	1.157	.999	1.340
	IPQ_EmotionalRep	.172	.063	7.555	1	.006	1.188	1.051	1.343
	COPE_SU	.730	.363	4.042	1	.044	2.075	1.019	4.228
	COPE_BD	.950	.564	2.833	1	.092	2.585	.855	7.812
	COPE_SB	-.100	.183	.296	1	.586	.905	.632	1.296
	Constant	-9.107	2.120	18.451	1	.000	.000		

a. Variable(s) entered on step 1: IPQ_Identity, IPQ_TimelineCyclical, IPQ_EmotionalRep, COPE_SU, COPE_BD, COPE_SB.

Casewise List[b]

Case	Selected Status[a]	Observed	Predicted	Predicted Group	Temporary Variable	
		HADS Anxiety Split into groups of Normal and Above Clinical			Resid	ZResid
67	S	N**	.856	A	-.856	-2.439
76	S	N**	.962	A	-.962	-5.004
84	S	N**	.913	A	-.913	-3.248
109	S	N**	.894	A	-.894	-2.905

a. S = Selected, U = Unselected cases, and ** = Misclassified cases.

b. Cases with studentized residuals greater than 2.000 are listed.

Appendix 13- Logistic Regression Analysis with Depression as Outcome SPSS Output

Case Processing Summary

Unweighted Cases[a]		N	Percent
Selected Cases	Included in Analysis	113	100.0
	Missing Cases	0	.0
	Total	113	100.0
Unselected Cases		0	.0
Total		113	100.0

a. If weight is in effect, see classification table for the total number of cases.

Dependent Variable Encoding

Original Value	Internal Value
Normal	0
Above Clinical	1

Block 0: Beginning Block

Iteration History[a,b,c]

Iteration		-2 Log likelihood	Coefficients
			Constant
Step 0	1	145.642	-.619
	2	145.630	-.640
	3	145.630	-.641

a. Constant is included in the model.

b. Initial -2 Log Likelihood: 145.630

c. Estimation terminated at iteration number 3 because parameter estimates changed by less than .001.

Classification Table[a,b]

	Observed		Predicted		
			HADS Depression Split into groups of normal and above clinical		Percentage Correct
			Normal	Above Clinical	
Step 0	HADS Depression Split nto groups of normal and above clinical	Normal	74	0	100.0
		Above Clinical	39	0	.0
	Overall Percentage				65.5

a. Constant is included in the model.

b. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 0	Constant	-.641	.198	10.478	1	.001	.527

Variables not in the Equation

			Score	df	Sig.
Step 0	Variables	IPQ_IllnessCoherence	8.749	1	.003
		IPQ_EmotionalRep	13.045	1	.000
		COPE_SU	11.895	1	.001
		COPE_ES	6.240	1	.012
		COPE_BD	9.531	1	.002
		COPE_SB	6.683	1	.010
	Overall Statistics		32.160	6	.000

Block 1: Method = Enter

Iteration History[a,b,c,d]

Iteration		-2 Log likelihood	Coefficients						
			Constant	IPQ_ Illness Coherence	IPQ_ Emotional Rep	COPE_ SU	COPE_ES	COPE_BD	COPE_SB
Step 1	1	112.485	-.387	-.114	.059	.315	-.205	.202	.130
	2	109.232	-.173	-.161	.082	.447	-.312	.217	.188
	3	109.092	-.136	-.171	.088	.485	-.345	.223	.202
	4	109.092	-.136	-.172	.088	.488	-.347	.223	.203
	5	109.092	-.136	-.172	.088	.488	-.347	.223	.203

a. Method: Enter

b. Constant is included in the model.

c. Initial -2 Log Likelihood: 145.630

d. Estimation terminated at iteration number 5 because parameter estimates changed by less than .001.

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	36.538	6	.000
	Block	36.538	6	.000
	Model	36.538	6	.000

Model Summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	109.092[a]	.276	.381

a. Estimation terminated at iteration number 5 because parameter estimates changed by less than .001.

Hosmer and Lemeshow Test

Step	Chi-square	df	Sig.
1	7.313	8	.503

Contingency Table for Hosmer and Lemeshow Test

		HADS Depression Split nto groups of normal and above clinical = Normal		HADS Depression Split nto groups of normal and above clinical = Above Clinical		Total
		Observed	Expected	Observed	Expected	
Step 1	1	10	10.570	1	.430	11
	2	11	10.001	0	.999	11
	3	9	9.580	2	1.420	11
	4	10	8.916	1	2.084	11
	5	9	8.433	2	2.567	11
	6	9	7.839	2	3.161	11
	7	4	7.025	7	3.975	11
	8	6	5.505	5	5.495	11
	9	4	4.060	7	6.940	11
	10	2	2.072	12	11.928	14

Classification Table[a]

	Observed		Predicted		
			HADS Depression Split nto groups of normal and above clinical		Percentage Correct
			Normal	Above Clinical	
Step 1	HADS Depression Split nto groups of normal and above clinical	Normal	65	9	87.8
		Above Clinical	17	22	56.4
	Overall Percentage				77.0

a. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I.for EXP(B)	
								Lower	Upper
Step 1[a]	IPQ_IllnessCoherence	-.172	.077	5.010	1	.025	.842	.724	.979
	IPQ_EmotionalRep	.088	.052	2.862	1	.091	1.092	.986	1.209
	COPE_SU	.488	.195	6.236	1	.013	1.628	1.111	2.387
	COPE_ES	-.347	.161	4.622	1	.032	.707	.515	.970
	COPE_BD	.223	.327	.467	1	.495	1.250	.659	2.372
	COPE_SB	.203	.168	1.460	1	.227	1.225	.881	1.702
	Constant	-.136	1.882	.005	1	.943	.873		

a. Variable(s) entered on step 1: IPQ_IllnessCoherence, IPQ_EmotionalRep, COPE_SU, COPE_ES, COPE_BD, COPE_SB.

Casewise List[b]

Case	Selected Status[a]	Observed	Predicted	Predicted Group	Temporary Variable	
		HADS Depression Split nto groups of normal and above clinical			Resid	ZResid
4	S	A**	.140	N	.860	2.475
12	S	A**	.137	N	.863	2.512
61	S	A**	.047	N	.953	4.518
105	S	N**	.951	A	-.951	-4.412

a. S = Selected, U = Unselected cases, and ** = Misclassified cases.

b. Cases with studentized residuals greater than 2.000 are listed.

Appendix 14- Logistic Regression Analysis with Positive Affect as Outcome SPSS Output

Case Processing Summary

Unweighted Cases[a]		N	Percent
Selected Cases	Included in Analysis	113	100.0
	Missing Cases	0	.0
	Total	113	100.0
Unselected Cases		0	.0
Total		113	100.0

a. If weight is in effect, see classification table for the total number of cases.

Dependent Variable Encoding

Original Value	Internal Value
Low	0
High	1

Block 0: Beginning Block

Iteration History[a,b,c]

Iteration		-2 Log likelihood	Coefficients
			Constant
Step 0	1	156.430	-.088
	2	156.430	-.089

a. Constant is included in the model.

b. Initial -2 Log Likelihood: 156.430

c. Estimation terminated at iteration number 2 because parameter estimates changed by less than .001.

Classification Table[a,b]

	Observed		Predicted		
			PANAS Positive Affect Group		Percentage Correct
			Low	High	
Step 0	PANAS Positive Affect Group	Low	59	0	100.0
		High	54	0	.0
	Overall Percentage				52.2

a. Constant is included in the model.

b. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 0	Constant	-.089	.188	.221	1	.638	.915

Variables not in the Equation

			Score	df	Sig.
Step 0	Variables	COPE_AC	18.788	1	.000
		COPE_SU	4.308	1	.038
		COPE_ES	8.314	1	.004
		COPE_BD	10.719	1	.001
		COPE_PR	19.595	1	.000
		COPE_Pla	9.823	1	.002
		COPE_Acc	9.891	1	.002
	Overall Statistics		38.077	7	.000

Block 1: Method = Enter

Iteration History[a,b,c,d]

Iteration		-2 Log likelihood	Coefficients							
			Constant	COPE AC	COPE SU	COPE ES	COPE BD	COPE PR	COPE Pla	COPE Acc
Step 1	1	114.105	-2.792	.258	-.167	.096	-.528	.255	.049	.126
	2	110.462	-3.690	.345	-.288	.173	-.741	.327	.062	.180
	3	110.263	-3.924	.364	-.329	.198	-.808	.348	.068	.197
	4	110.262	-3.939	.365	-.332	.200	-.814	.350	.069	.198
	5	110.262	-3.939	.365	-.332	.200	-.814	.350	.069	.198

a. Method: Enter

b. Constant is included in the model.

c. Initial -2 Log Likelihood: 156.430

d. Estimation terminated at iteration number 5 because parameter estimates changed by less than .001.

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	46.168	7	.000
	Block	46.168	7	.000
	Model	46.168	7	.000

Model Summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	110.262[a]	.335	.447

a. Estimation terminated at iteration number 5 because parameter estimates changed by less than .001.

Hosmer and Lemeshow Test

Step	Chi-square	df	Sig.
1	10.049	8	.262

Contingency Table for Hosmer and Lemeshow Test

		PANAS Positive Affect Group = Low		PANAS Positive Affect Group = High		Total
		Observed	Expected	Observed	Expected	
Step 1	1	11	10.606	0	.394	11
	2	8	9.697	3	1.303	11
	3	8	8.892	3	2.108	11
	4	7	7.682	4	3.318	11
	5	8	6.469	3	4.531	11
	6	8	5.418	3	5.582	11
	7	6	4.321	5	6.679	11
	8	2	3.004	9	7.996	11
	9	1	1.825	10	9.175	11
	10	0	1.086	14	12.914	14

Classification Table[a]

	Observed		Predicted		
			PANAS Positive Affect Group		Percentage Correct
			Low	High	
Step 1	PANAS Positive Affect Group	Low	45	14	76.3
		High	15	39	72.2
	Overall Percentage				74.3

a. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I.for EXP(B)	
								Lower	Upper
Step 1[a]	COPE_AC	.365	.162	5.083	1	.024	1.440	1.049	1.978
	COPE_SU	-.332	.207	2.573	1	.109	.717	.478	1.077
	COPE_ES	.200	.161	1.554	1	.213	1.222	.892	1.674
	COPE_BD	-.814	.389	4.365	1	.037	.443	.207	.951
	COPE_PR	.350	.151	5.403	1	.020	1.419	1.056	1.906
	COPE_Pla	.069	.173	.158	1	.691	1.071	.764	1.502
	COPE_Acc	.198	.187	1.124	1	.289	1.219	.845	1.760
	Constant	-3.939	1.760	5.011	1	.025	.019		

a. Variable(s) entered on step 1: COPE_AC, COPE_SU, COPE_ES, COPE_BD, COPE_PR, COPE_Pla, COPE_Acc.

Casewise List[b]

Case	Selected Status[a]	Observed	Predicted	Predicted Group	Temporary Variable	
		PANAS Positive Affect Group			Resid	ZResid
3	S	H**	.107	L	.893	2.892
35	S	H**	.112	L	.888	2.822
84	S	H**	.116	L	.884	2.764

Appendix 15- Logistic Regression Analysis with Negative Affect as Outcome SPSS Output

Logistic Regression

Case Processing Summary

Unweighted Cases[a]		N	Percent
Selected Cases	Included in Analysis	113	100.0
	Missing Cases	0	.0
	Total	113	100.0
Unselected Cases		0	.0
Total		113	100.0

a. If weight is in effect, see classification table for the total number of cases.

Dependent Variable Encoding

Original Value	Internal Value
Low	0
High	1

Block 0: Beginning Block

Iteration History[a,b,c]

Iteration		-2 Log likelihood	Coefficients
			Constant
Step 0	1	155.152	-.230
	2	155.152	-.231
	3	155.152	-.231

a. Constant is included in the model.

b. Initial -2 Log Likelihood: 155.152

c. Estimation terminated at iteration number 3 because parameter estimates changed by less than .001.

Classification Table[a,b]

	Observed		Predicted		
			PANAS Negative Scale Group		Percentage Correct
			Low	High	
Step 0	PANAS Negative Scale Group	Low	63	0	100.0
		High	50	0	.0
	Overall Percentage				55.8

a. Constant is included in the model.

b. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)
Step 0	Constant	-.231	.189	1.489	1	.222	.794

Variables not in the Equation

			Score	df	Sig.
Step 0	Variables	IPQ_EmotionalRep	21.858	1	.000
		COPE_SD	9.176	1	.002
		COPE_De	4.515	1	.034
		COPE_SU	10.937	1	.001
		COPE_BD	15.069	1	.000
		COPE_Ve	12.118	1	.000
		COPE_SB	8.749	1	.003
	Overall Statistics		33.067	7	.000

Block 1: Method = Enter

Iteration History[a,b,c,d]

Iteration	-2 Log likelihood	Coefficients							
		Constant	IPQ_EmotionalRep	COPE_SD	COPE_De	COPE_SU	COPE_BD	COPE_Ve	COPE_SB
Step 1	118.725	-5.009	.111	.076	.058	.176	.371	.170	.063
	115.666	-6.922	.143	.097	.112	.279	.504	.277	.092
	115.535	-7.411	.150	.103	.126	.309	.544	.303	.102
	115.535	-7.437	.150	.103	.127	.310	.547	.304	.103
	115.535	-7.437	.150	.103	.127	.310	.547	.304	.103

a. Method: Enter

b. Constant is included in the model.

c. Initial -2 Log Likelihood: 155.152

d. Estimation terminated at iteration number 5 because parameter estimates changed by less than .001.

Omnibus Tests of Model Coefficients

		Chi-square	df	Sig.
Step 1	Step	39.617	7	.000
	Block	39.617	7	.000
	Model	39.617	7	.000

Model Summary

Step	-2 Log likelihood	Cox & Snell R Square	Nagelkerke R Square
1	115.535[a]	.296	.396

a. Estimation terminated at iteration number 5 because parameter estimates changed by less than .001.

Hosmer and Lemeshow Test

Step	Chi-square	df	Sig.
1	6.182	8	.627

Contingency Table for Hosmer and Lemeshow Test

		PANAS Negative Scale Group = Low		PANAS Negative Scale Group = High		Total
		Observed	Expected	Observed	Expected	
Step 1	1	11	10.252	0	.748	11
	2	10	9.471	1	1.529	11
	3	9	8.877	2	2.123	11
	4	6	8.052	5	2.948	11
	5	8	7.240	3	3.760	11
	6	5	6.367	6	4.633	11
	7	5	5.100	6	5.900	11
	8	6	3.771	5	7.229	11
	9	2	2.700	9	8.300	11
	10	1	1.171	13	12.829	14

Classification Table[a]

	Observed		Predicted		
			PANAS Negative Scale Group		Percentage Correct
			Low	High	
Step 1	PANAS Negative Scale Group	Low	50	13	79.4
		High	18	32	64.0
	Overall Percentage				72.6

a. The cut value is .500

Variables in the Equation

		B	S.E.	Wald	df	Sig.	Exp(B)	95% C.I.for EXP(B)	
								Lower	Upper
Step 1[a]	IPQ_EmotionalRep	.150	.054	7.787	1	.005	1.162	1.046	1.291
	COPE_SD	.103	.230	.201	1	.654	1.109	.707	1.739
	COPE_De	.127	.417	.092	1	.761	1.135	.501	2.570
	COPE_SU	.310	.188	2.723	1	.099	1.364	.943	1.972
	COPE_BD	.547	.396	1.911	1	.167	1.728	.796	3.752
	COPE_Ve	.304	.250	1.477	1	.224	1.356	.830	2.214
	COPE_SB	.103	.153	.451	1	.502	1.108	.821	1.495
	Constant	-7.437	1.565	22.596	1	.000	.001		

a. Variable(s) entered on step 1: IPQ_EmotionalRep, COPE_SD, COPE_De, COPE_SU, COPE_BD, COPE_Ve, COPE_SB.

Casewise List[b]

Case	Selected Status[a]	Observed	Predicted	Predicted Group	Temporary Variable	
		PANAS Negative Scale Group			Resid	ZResid
67	S	L**	.809	H	-.809	-2.059
77	S	L**	.917	H	-.917	-3.325

a. S = Selected, U = Unselected cases, and ** = Misclassified cases.

b. Cases with studentized residuals greater than 2.000 are listed.

Appendix 16- Notes to Authors: Research in Developmental Disabilities

Appendix 17- Notes to Authors: Journal of Cognitive and Behavioural Psychotherapy