STRESS AND HEALTH-RELATED QUALITY OF LIFE IN MOTHERS SEEKING A DIAGNOSIS OF HIGH-FUNCTIONING AUTISM FOR THEIR CHILD, A MIXED METHODS INVESTIGATION

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ABSTRACT

Objective: To investigate mothers' experiences in raising a child with suspected high functioning autism or Asperger's syndrome while trying to get a diagnosis for their child, specifically focusing on stress and health-related quality of life. Mothers who already have a diagnosis for their child acted as a comparison group.

Method: A cross-sectional mixed methods approach was applied. The Parenting Stress Index Short Form (PSI/SF), the 12 Item Short-Form Health Survey (SF12) and a demographic questionnaire was administered to a convenience sample of 40 mothers in the experimental group and 44 in the comparison group, all of whom resided in the UK. Qualitative semi-structured interviews with a sample of 4 mothers in each group followed to expand on those findings; data was analysed using thematic analysis.

Results: Although both groups displayed clinically high mean scores for all the PSI/SF subscales and overall stress, the experimental group displayed significantly higher scores in the Parenting Stress and Difficult Child subscales and overall stress scores. The experimental group scored significantly worse in the Mental Component Summary scores of the Health Survey than the comparison group, indicating poorer mental-health related quality of life. The interviews, which discussed mother's experiences of raising their child, the diagnostic process and its implications, also supported the quantitative findings.

Implications: This study highlights the health and mental health implications on the large population of mothers seeking diagnoses of autism for their children. This population group is largely neglected in the literature. This brings into focus areas where further in-depth research and development of support systems for this population would potentially benefit these parents, specifically in terms of health related quality of life and it supports the drive by the National Autistic Society for earlier diagnoses and more efficient diagnostic processes.

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Chapter 1

Research: Stress and Health-Related Quality of Life in Mothers Seeking a Diagnosis of High-Functioning Autism for Their Child. A Mixed Methods Investigation.

INTRODUCTION

This literature review looks at high functioning autism and Asperger's syndrome and the challenges and health consequences mothers face when raising a child with this condition. Parental stress and quality of life is investigated with particular emphasis on the challenges and considerations of getting a diagnosis for their children.

Autism and Asperger's syndrome are classified as pervasive developmental disorders that are distinguished by a number of symptoms rather than a single symptom (American Psychiatric Association, 2000). It is no longer considered a rare disorder as recent statistics have found it to be more prevalent in children than cancer, diabetes, spina bifida, and Down syndrome (Kishore and Basu, 2011).

The Diagnosis

There is a variation in the estimated prevalence of Autism Spectrum Disorder (ASD) in the UK. According to the National Autistic Society autism can affect an estimated 60 of every 10,000 people under 8 years old in the UK (National Initiative for Autism Screening and Assessment (NIASA), 2003). A study based at the South Thames Region produced an estimated prevalence of 116 per 10,000 children aged 9-10 by reviewing children on the Special Educational Needs register, this is approximately 1% of the child population in the UK (Baird *et al,* 2006). A more recent study reviewed current numbers of children diagnosed, those with Special Educational Needs and then included a direct school population screening to gain an estimate of diagnosed and non-diagnosed children between 5 and 9 years old within the Cambridge area. These researchers estimated that there are approximately 157 children per 10,000 with Autism, forty per cent of whom are undiagnosed, in the UK population (Baron-Cohen *et al,* 2009), this is 2.5 times the National Autistic Society estimation (Baird *et al,* 2006).

There are two standardised classification systems that are used to diagnose autism; the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR) (American Psychiatric Association, 2000); and the International Classification of Diseases (ICD-10) (World Health Organisation, 1992). Both list the following criteria for a diagnosis of autism:

- A restriction of reciprocal social interaction

- A restriction of reciprocal communication

- A restriction of imagination as reflected in a restricted range of behaviours.

These 3 impairments together are referred to as the 'Triad' and they must be present in the first three years of a child's life in order for a correct diagnosis to be made (Peeters, 1997), this will include a delay or abnormal functioning in social interaction, language or imaginative play (World Health Organisation, 1992). It is possible for autism to be diagnosed as young as 14 months old, however diagnosis is likely to be more stable when the child is over 3 years old (Landa, 2008). People with autism generally have some impairment within each of the three categories, although severity of each symptom may vary (Peeters, 1997) and speech deficits, for example language delays, are the most common symptom first raised by parents in children between the ages of 1 and 5 years (Filipek *et al,* 1999). When a child has a language delay it means they are not using single words by two years of age, and/or phrase speech by three years of age (Baron-Cohen *et al,* 2001).

The National Institute of Clinical Excellence outlined guidance on how the diagnostic process should be carried out in the UK (National Institute of Clinical Excellence, 2011). Once concerns have been raised about a child, he or she should be referred to an autism team and be assigned an autism case coordinator. This case coordinator should be a single point of contact for the parents or carers and should give them any relevant information regarding diagnostic assessment and address any concerns raised by the parent/carer. The autism team should include or have access to the following professionals: paediatrician or paediatric neurologist, child and adolescent psychiatrist, educational psychologist, clinical psychologist and occupational therapist.

The diagnostic assessment should be based on parental concerns, child's experiences, a developmental and medical history, assessment of social communicational skills and behaviours in relation to the ICD-10 or DSM-IV, a physical examination and a development of a profile of the child's strengths, skills, impairments or needs. Parents should be informed of the findings of this assessment in person, without delay. Parents/carers should also be given information on what autism is and how likely autism is likely to affect the child's development and function. If there is continued uncertainty about the results of the assessment, by a parent/carer or practitioner then a second opinion should be obtained. Once the diagnosis has been communicated a follow-up appointment should be offered, 6 weeks following the assessment for further discussion.

Parents are usually the first to notice unusual behaviours in their child (Filipek *et al,* 1999) and this is usually before the child turns 3 years old (Jónsdóttir *et al,* 2011). If the child's problems are severe the diagnosis is likely to be made quite early (Shattuck *et al,* 2009). Autism specialists use a variety of methods to identify the disorder. A tool commonly used by specialists in the United Kingdom is the Autism Diagnostic Observation Schedule which is a group of structured tests, tailored to different age groups which enables the observer to evaluate the child's language and social behaviour (Akshoomoff, Corsello and Schmidt, 2006). A structured interview is also used to elicit information from parents about the child's behaviour and early development such as the Autism Diagnostic Interview-Revised (Gotham *et al,* 2008).

A child with Asperger Syndrome (AS) will meet the same criteria for a diagnosis of autism but will have no history of cognitive or language delay (World Health Organisation, 1992). According to the National Autistic Society (2003) Asperger syndrome has become shorthand for those within the autism spectrum who have good structural language skills and no general intellectual impairment, i.e. high functioning autism (National Initiative for Autism Screening and Assessment (NIASA), 2003). Witwer et al (2008) reviewed studies assessing the validity of Autism Spectrum Disorder Subtypes including Asperger's Syndrome and High Functioning Autism. Overall they found evidence to support the different categories of autism and Asperger's syndrome, although the differences were found in

IQ levels. Autism can occur at all levels of abilities, whereas Asperger's syndrome is often described as having high functioning autism as they will have average or above average IQ levels (Fitzgerald and Corvin, 2001). Therefore current research data does not support the separation of Asperger syndrome and the high functioning autism as distinct disorders (Fitzgerald and Corvin, 2001). However Fitzgerald et al (2001) suggest that the term Asperger's syndrome is more acceptable to parents with children who have higher-functioning autism as despite relatively normal cognitive ability they will still have comprehensive difficulties.

High functioning autism is a condition that can present huge problems for parents as they not only have to cope with the demands of raising a child with a disability but there are often social consequences that accompany this condition (Gray, 2002a). It can come with impaired social relations, obsessions, uneven levels of intellectual and cognitive functioning and peculiarities in language (Gray, 2002a). This condition will have disabilities which are not immediately obvious to the public and the low visibility of the symptoms and the almost normal social involvement of the children means that they and their families are exposed to more incidents of social stigma without the obvious explanation of their condition (Gray, 2002a).

A survey of 770 families found that on average children with autism do not receive a diagnosis until around 5.5 years, and children with Asperger's syndrome do not receive a diagnosis until around the age of 11 (Howlin and Asgharian, 1999). As a result the National Autistic Society, the Royal College of Psychiatrists and the Royal College of Paediatrics and Child Health have collaborated to produce the National Autistic Plan for Children (National Initiative for Autism Screening and Assessment (NIASA), 2003, Montes and Halterman, 2007). This is a set of guidelines encouraging a more efficient diagnostic process in all stages including identification, assessment diagnosis, and access to early interventions. It also highlights the need for early intervention for the child and family and for more and better skilled practitioners. The National Autism Plan recommends that diagnosis is completed 30 weeks from first concern, however in most cases a diagnosis takes much longer (Dover and Couteur, 2007).

In a comparison study looking at characteristics of children diagnosed before and after aged 6 Jónsdóttir et al (2011) found that children with a lower IQ, low verbal status and a history of autistic regression are more likely to receive a diagnosis of childhood autism before they are 6 years old, which can explain why those children who have high functioning autism are usually not diagnosed until 11 years of age. Half of the children who had received a diagnosis after the age of 6 had received other developmental diagnoses prior to the ASD diagnosis. This particular research found that there was no difference between the group who were diagnosed early and the group who were diagnosed later in the numbers of parents who were first concerned and raised their concern before their child turned 3 years old.

Many reasons have been cited as to why there are delays in diagnosis for children with high functioning autism. For example, children's psychological problems are more unpredictable than those of adults and can change dramatically as the child grows older. Children also cannot communicate their problems as easily as adults can and sometimes this results in frustrations being displayed in disruptive behaviours, with this other abnormal behaviours can be missed or ignored (Peeters, 1997). Certain behaviours can be specific to particular situations or environments that are causing the child distress and the parents and carers will then have conflicting perceptions of the child's behaviour (Peeters, 1997).

Symptoms of Asperger's syndrome or high functioning autism can often be misinterpreted and wrongly diagnosed under other pervasive developmental disorders such as schizophrenia, Rett disorder, multiple complex developmental disorder and attention deficit hyperactivity disorder (Fitzgerald and Corvin, 2001). Misdiagnosis can lead to further problems for the child, for example the child could be given incorrect prescriptions such as neuroleptics or psychostimulants which will have their own side effects. This confusion in the diagnosis and its process often leads to families seeking unhelpful therapies or joining the wrong support groups (Fitzgerald and Corvin, 2001).

The Meaning of Diagnosis for Parents

Not many childhood conditions are comparable to high functioning autism as symptoms are usually more visible, therefore easier to diagnose, or they do not come with the behavioural symptoms that can lead to stigma on the mother's parenting skills (Lee *et al*, 2009). However, a literature search was conducted to investigate the meaning of a diagnosis in childhood disorders, diseases and conditions, including medically unexplained conditions, in relation to parental stress and quality of life, only a few studies were found. In one condition related to high parental stress, attention deficit hyperactivity disorder (ADHD), it was found that the child's behavioural symptoms associated with this condition were perceived by onlookers to be caused by a conflict in the parent-child relationship, such as negative interactions or controlling behaviour in the parent (Fletcher *et al,* 1996). Although a diagnosis might reduce this stigma, no studies have looked into how a diagnosis might help a parent to cope with this. Another study found that for ADHD it was not the diagnosis but how the parents interpreted their child's symptoms which affected the level of stress they experienced as a result of their child's behaviours (Whalen *et al*, 2011), for example, if they viewed their child's behaviours as talents or quirks they appraised their experiences in raising their child as less stressful as those who viewed their children's behaviours as difficult.

Makela *et al* (2009) looked at parental perceived value of a diagnosis of intellectual disability by comparing interviews of those with a diagnosis to those without a diagnosis. They found that parents felt the need for a diagnosis most strongly when they first noted concerns and this diminished over time. However they noticed no differences between the group's experiences with regards to the diagnosis. No studies could be found that investigated the meaning of a diagnosis for a child on parental stress directly.

A diagnosis can have some impact on a parent's health seeking behaviour. For example, one study found that families with a child diagnosed with epilepsy sometimes compromised their quality of life to avoid the stigma attached to the diagnosis by withdrawing from social activities or not seeking help (Austin *et al*, 2004). Another study reviewing health seeking behaviours found that families of children with emotional disorders often felt that people in their community would blame them for

their child's condition and marginalise them if they were to go to mental health services thus affecting their choices of whether to utilise these services (Brannan & Heflinger, 2006). Therefore knowledge of their child's condition and the reluctance to seek appropriate help may have an impact on a parent's stress levels and health-related quality of life, however the extent of this has not yet been quantified by published research.

Gaining a diagnosis of autism

Jim Sinclair, the Autism Network International co-ordinator once reported that parents often found that learning their child is autistic is the most traumatic period in their life (Sinclair, 1993). They experience grief for the normal child they expected and wanted to have and parents must learn to adjust to the realisation that the relationship they've been looking forward to isn't going to happen (Sinclair, 1993). However, in a pilot qualitative study involving 4 families residing in Wales, Midence and O'Neill (1999) found that parents expressed feelings of relief when a correct diagnosis of autism was finally made. This helped them to understand and accept autism and the limitations this imposed on their child, to help the family adapt to the condition and to foster more realistic expectations of their child's future. They also found that prior to receiving a diagnosis the parents expressed difficulties and confusions in understanding their child's behaviours (Midence and O'Neill, 1999).

Although it has been found that families customarily express the desire to be informed as soon as possible about their child's condition, children with autism can often receive no diagnosis or an inaccurate diagnosis (Filipek *et al,* 1999). Practitioners often hesitate to discuss the possibility of a autism diagnosis with parents of young children displaying symptoms because they are concerned it will lead to family distress, they are not confident in their diagnosis or they hope that symptoms will reverse over time (Filipek *et al,* 1999). Some are concerned about the implications of labelling a child (Filipek *et al,* 1999).

Rose (2011) looked at the parent's experiences of the diagnostic process for autism spectrum disorders and parental stress and satisfaction regarding their experiences. She found that approximately 20% of parents interviewed reported that their child's health professional dismissed their initial concerns and the average length of time to receive a diagnosis was nearly two years (Rose, 2011). Nearly 80% of the parents interviewed reported experiencing high or very high stress due to diagnostic process for their child and one third were dissatisfied with the diagnostic process. Rose (2011) also found the child's age at diagnosis and the length of the diagnostic process did not significantly correlate with parent satisfaction, but Sivberg (2002) found that the longer length of time increased parental stress. Keenen et al (2009) also found that the autism diagnostic and planning processes 'extremely stressful' for parents. They concluded that this was because the statutory diagnosis takes such a long time and the care and education plans do not allow for full parental participation (Keenan *et al,* 2010).

The failure of healthcare workers to provide a quick diagnosis can have several negative effects on mothers; firstly it can be difficult for mothers to reiterate the information from the healthcare worker to their husbands as the information can be complicated and often inaccurate in the early stages of the referral experience (Gray, 2002a). Secondly, the inaccurate diagnosis can often expose the mother to accusations of bad parenthood by others, including healthcare workers and family members. This can be particularly stressful when no other explanation is provided to account for the child's behaviour (Gray, 2002a).

Even with a diagnosis it can sometimes be difficult to gain recognition for the challenges mothers face when raising a child with autism, for example, husbands and grandparents can be highly critical of the mother's child raising skills and will often deny that the child has a disability (Gray, 2002b, Gray, 2003). The accusations of bad parenting usually decline however after the diagnosis, although mothers often still feel a considerable amount of guilt and depression over their child's disability (Gray, 2003). Nissenbaum et al (2002) found in interviews conducted on 17 parents after receiving a diagnosis for their child that parents sometimes experienced negative emotions such as denial, emotionality and misperceptions of the diagnosis. Mostly however they found that parents

expressed relief because they no longer felt they were to blame and they had an explanation for themselves and their child's behaviour (Nissenbaum, Tollefson and Reese, 2002).

Once a diagnosis is made a decision regarding appropriate support services can be made, parents can learn how to best care for and manage their child (Goin and Myers, 2004). With an earlier diagnosis parents can educate themselves about their child's condition and ease long-term family stress through the knowledge of what is affecting their child and how to manage it (Goin and Myers, 2004).

Timing of the diagnosis

Autism Spectrum Disorder has been described as quite heterogeneous, it can be very debilitating and will have a lifelong course. The general consensus among researchers therefore, is that intervention should start at a very early age as a means of enhancing prognosis (Matson, Rieske and Tureck, 2011) .

When children with high functioning autism go undiagnosed for years it can present many difficulties for that child. For example, these children would usually attend school with children who do not have autism and will continuously struggle with the demands of their education without their needs supported (Filipek *et al,* 1999). In adulthood whether or not people with this condition receive a diagnosis, they will still be expected to work, live independently and fulfil the typical adult role (Gray, 2002a). These difficulties can also cause great stress for their families, who will recognise the child's challenges but have difficulty convincing others that their child has a disability (Filipek *et al,* 1999).

The timing of the diagnosis for their child is an important issue for parents and their reactions; parents report higher stress levels if the intervention is received late in the child's life (Hastings and Johnson, 2001). Late diagnosis can lead to uncertainty, confusion, hostility, and in some cases

avoidance of the child (Wiggins, Baio and Rice, 2006). It can also lead to a delay in accessing behavioural interventions, appropriate social support systems and can lead to parents unwittingly reinforcing problem behaviours (Keenan *et al,* 2010). The first and primary problem as reported by parents raising a child with autism is the amount of effort that can be required to get a proper diagnosis of autism for their child and then to locate an intervention or treatment program that is both adequate and accessible for the children and for the families (Sivberg, 2002).

The advantages of an early diagnosis include earlier educational planning and treatment and delivery of any appropriate medical care for the child (Filipek *et al,* 1999). Early intervention results in improved outcomes in most young children with autism with 75% or more demonstrating significant increases in rates of developmental progress and intellectual performance (Rogers, 1998), particularly for children who receive intervention before or during pre-school years (Ozonoff and Cathcart, 1998).

Stress

Parents often use the terms "stressful" and "on-going" when talking about their experiences raising a child with autism as they often do not get any respite from the continuous care they have to provide to their child with special needs (Hutton and Caron, 2005, Rao and Beidel, 2009, Portway and Johnson, 2005b). Lazarus and Folkman (1984) defined psychological stress as a particular relationship between the person and the environment that is appraised by the person as taxing or exceeding on his or her resources and endangering his or her welfare. They proposed a transactional model of stress which describes how it is the interpretation or cognitive appraisal of a stressful situation rather than the actual stress itself which is important to understand. Cognitive appraisal is a mental process by which an individual will assess whether or not a stressor is threatening, challenging or potentially harmful and whether or not he/she means to deal with that stressor. The first factor, primary appraisal is where the individual will look at the situation or stimulus and how it affects his wellbeing. If the situation or stimulus is seen as threatening, harmful or challenging then a

secondary appraisal is made. The secondary appraisal is where the individual evaluates whether he has the resources to deal with the demand (Lazarus and Folkman, 1984) by considering factors such as: what options are available and what is the likelihood that these options will be successful in reducing stress. Reappraisal occurs when we revaluate the stressful situation and our responses to it as they change. This conceptualisation of stress is still being applied within health psychology research to understand stress and its effects today, for example, González-Ramírez, García-Campayo and Landero-Hernández (2011), Radat and Koleck (2011), Kim and So (2012), Lindfors, Boman and Alexanderson (2012), and Del-Pino-Casado *et al* (2011).

Studies which have compared a parent's life with a child with autism to that of a parent with a child with mental retardation or psychological disorder found that those raising a child with ASD reported higher levels of stress, depression, anxiety, somatic complaints and burnout (Rao and Beidel, 2009 and Weiss, 2002).

Mothers of children with autism are significantly more likely to report that their child was 'much harder to care for than other children his or her age' and that 'they have given up more of their life than expected in the previous month' than parents with children who do not have autism (Montes and Halterman, 2007). Bromley et al (2004) who interviewed 68 mothers of children with ASD residing in Manchester found that over half reported serious psychological distress (Bromley *et al,* 2004).

There are many stressors associated with raising a child with autism, for example, fits of temper, specific clothes obsession, need for sameness, defiance, sleeping problems, phobia of crowded places (Akshoomoff and Stahmer, 2006). Mothers find that their stress levels increase when their child embarrasses them with their unreasonable crying, giggling, or peculiar behaviours in public places (Hall and Graff, 2011b).

Hutton & Caron (2005) found that family life is affected when raising a child with autism and factors such as less time for family activities, lack of flexibility or spontaneity and pressures on the marital relationship add to the stress felt by mothers raising a child with autism. Another study found that

making restrictions on family life were sometimes used as a method of avoiding possible embarrassment from their child's problem behaviour, such as becoming socially reclusive. This and the parental stress reduced in time in families where the child did not have severe behavioural difficulties, it however, increased in time where the child's behavioural difficulties remained and the child became harder to control (Gray, 2002b). Parents views of the significance of their daily stressors such as communicational difficulties with their child and managing their child's behaviour reduced over time (Gray, 2002b). Gray (2002) suggests that this could be due to either an improvement in the child's behaviours and abilities or the parent's capacity to understand their child more and manage their expectations.

Rao & Beidle found that the higher intellectual functioning of children with high functioning autism did not compensate for the stress associated with parenting children with ASD (Rao and Beidel, 2009). Children with high functioning autism have abilities that often hide their significant social struggles, parents are frequently denied the acknowledgement of the support they need and struggle to get the services need for their children (Tsatsanis, Foley and Donehower, 2004).

Stigma

Parents of children with ASD can face social stigma, which often leads to social rejection (Gray, 2002b). Gray (2002) found that parents felt that others were critical of their child-raising abilities, not accepting of them and made them feel embarrassed. This was most commonly felt in public situations like shopping and social outings. Of the participants whom Gray (2002) interviewed in his study of stigma and ASD about half had experienced avoidance, overtly hostile staring or rude comments from strangers, friends or acquaintances. Having a diagnosis allows these parents to sometimes diffuse a situation, by relaying the information of the diagnosis to the stranger who stares or give rude comments, leading them to be embarrassed or apologetic (Gray, 2002a). However, in some occasions this does not diffuse the situation as either information is ignored by

the aggressor or the parent chooses not to relay the information out of respect for their child and his or her aspirations to be normal (Gray, 2002a).

Mothers are much more likely to experience the stigma than fathers (Gray, 2002a). Gray (2002) suggests that the reasons for this is are that mothers are more likely to engage in external social situations such as shopping with their children. Mothers may also be attributed more responsibility than their husbands for their child's behaviour. It is often the mother who takes on the majority of stresses and worries of raising a child with autism. Gray (2003) found in interviews with 32 mothers and 21 fathers that although the fathers noted the severe difficulties their child's autism presented for their families, it was generally the mothers who were likely to feel the impact of their child's autism. Mothers found that having a child with autism disrupted their careers, violated their expectations of domesticity, brought them into conflict with outside agencies and increased their exposure to negative social reactions (Gray 2002). Mothers were more likely to be blamed and to blame themselves for their child's problems and have their identities threatened by the illness and disability of their children (Sharpley, Bitsika and Efremidis, 1997); (Gray, 2003). According to Sharpley et al (1997) the three most stressful factors associated with raising a child with autism are: concern over the permanency of the condition, poor acceptance of the autistic behaviours by society and family members and the very low levels of social support received by parents.

The consequences, therefore, of raising a child with high functioning autism can mean mothers experience high levels of stress for a variety of reasons, as mentioned above. This can also have an impact on health-related quality of life.

Health-Related Quality of life

Lazarus and Folkman wrote that the importance of the appraisal and coping processes in their cognitive-appraisal model is that they affect the adaptational outcomes which are: 1) functioning in work and social living; 2) morale and life satisfaction; 3) somatic health (Lazarus and Folkman, 1984). Frank-Stromborg (1988) wrote that quality of life is the equivalent of psychosocial constructs,

including life satisfaction, happiness, mental well-being, adjustment, functional status, and health status. These can be influenced by one's self-perceptions i.e., the self-perceived position in life and how that position aligns with the individual's goals and expectations (Lee *et al*, 2009). Quality of life therefore is affected by physical health factors and mental wellbeing, as well as the perception of life circumstances (Lee *et al*, 2009). Parents of children with developmental disabilities are more vulnerable to lower quality of life than those whose children who do not have a developmental disability (Portway and Johnson, 2005a). Portway (2005) suggests that this is because they are confronted with daily and often lifelong challenges. For example, their child is likely to have a dependency on them for lifelong social and financial support.

Parenting a child with ASD can impact on health, for example a study found that mothers with children with Asperger's syndrome or high functioning autism indicated significantly poorer health-related quality of life than the controls. They also reported a relationship between maternal well-being and the child's behavioural characteristics, but not in paternal well-being (Allik, Larsson and Smedje, 2006). Other researchers looked at the impact of children's symptom severity and parenting stress on the psychological adjustment of 68 parents of children with ASDs, they concluded that parenting stress and children's symptom severity predicted parental depression (Benson, 2006). A more recent study found that lower physical health in mothers of children with ASD was linked to what they called higher caregiving stress (Johnson *et al*, 2011).

Research looking into the physical health of parents raising a child with ASD is limited; research tends to focus on the mental health of a caregiver. However there has been some research which looks at the health-related quality of life of caregivers of children with disabilities. For example Murphy et al (2007) noted that 41% of participants interviewed about their long-term care for their child with severe disabilities reported that their health had worsened over the past year (Murphy *et al*, 2007). The researchers attributed this to a lack of time to recuperate from providing care for their children and decreasing energy. Lee et al (2009) looked at both the physical and mental health-related quality of life of 89 parents of children with high-functioning autism spectrum disorders and compared them to 46 parents of children without disabilities. They found that stress, income and

the number of children contributed to variance in physical health-related quality of life. They also found that demographics and psycho-social variables accounted for low mental health-related quality of life, particularly income and stress (Lee *et al,* 2009).

Socio Economic Status

It is possible that socioeconomic status can play a part in whether a child is diagnosed or not and how a parent experiences stress and quality of life, however the evidence is mixed. When reviewing diagnosis, one study compared median household income data between 4 counties in the USA and found that the prevalence of diagnosed autism was highest in the wealthiest county (Thomas *et al*, 2012). Fountain *et al* (2011) looked at the age of diagnosis and its links with parental poverty and education. They found that children of those parents who were less educated and more poverty stricken were diagnosed at a later age. However, in a cohort study based in Denmark, parental socioeconomic was measured and the child's diagnostic status was compared 27 years later, they found no association (Larsson *et al*, 2005).

Several studies have found that women who are below the poverty line are more likely to experience depression and other mental health disorders (Groh, 2007; Belle & Doucet, 2003; Pirinicci *et al*, 2008) and this is because they are less likely to have access to social support (Pirinicci *et al,* 2008; Bell & Doucet, 2003). This may be exacerbated in the population of mothers raising a child of autism and has not yet been explored.

Systematic Review

In a systematic search of the literature (using Medline 1966–November 2011, Embase 1980-November 2011, PsycInfo 1974 – November 2011, Science Direct, Google and CINAHL Plus) a keyword search was conducted to find previous studies looking at the effects of non-diagnosis on parental stress and quality of life. Some studies highlighted that the length of time taken to receive a diagnosis of autism still takes a long time and can occur at a late age for children with high functioning autism (Jónsdóttir *et al,* 2011, Rose, 2011), while often these children receive wrong

diagnoses before learning their true diagnosis (Fitzgerald and Corvin, 2001). A few studies discuss the stress parents, mothers in particular, experience in correlation with the length of time taken for diagnosis (Keenan *et al,* 2010, Sivberg, 2002, Mulimba, 2002), and the relief they feel when the diagnosis is made (Midence and O'Neill, 1999, Nissenbaum, Tollefson and Reese, 2002). However these were retrospective studies and no studies were found that compared the stress felt in parents who do not have a diagnosis for their child to those who do.

There were a limited number of studies which investigated quality of life in parents who raise a child with an autism diagnosis (Portway and Johnson, 2005b, Lee *et al,* 2009, Allik, Larsson and Smedje, 2006, Benson, 2006, Johnson *et al,* 2011, Murphy *et al,* 2007). Yet no studies were found which looked at quality of life in parents of children who do not have a diagnosis, prior to their diagnosis or while they are in the process to receiving a diagnosis.

Summary

The Transactional model of stress illustrates how an individual will assess whether or not a stressor is threatening, challenging or potentially harmful and whether or not he/she has the means to deal with that stressor. The outcomes of this cognitive process will affect the stress levels experienced by that individual and their health related quality of life (or adaptational outcomes). Raising a child with high functioning autism is often seen as on-going and stressful and some of the coping methods available to parents who have a diagnosis for their child will not be available to parents who do not have a diagnosis. For example, parents who receive a diagnosis describe being able to seek appropriate support, experience acceptance and have an increased understanding of autism and what that means for their child and having an explanation to diffuse accusations of bad parenting as things that have helped them deal with their daily stresses. Those mothers who do not yet have a diagnosis for their child have the additional stressor of going through the diagnostic process. No studies have looked at the stresses and health related quality of life of mothers who are seeking a

diagnosis for their child, except in retrospect, and none have compared the stress and health related

quality of life to those mothers who have a diagnosis for their child.

RESEARCH AIMS

The aim of this study is to investigate mothers' experiences in raising a child with suspected high functioning autism or Asperger's syndrome while trying to get a diagnosis for their child. Specifically the study will focus on mothers' stress and quality of life. Mothers who already have a diagnosis for their child will act as a comparison group. This study will measure these factors using questionnaires and semi-structured interviews. The mixed methods approach has been chosen to allow for investigation into potential statistical differences between a representative sample and to expand on these with a more in-depth investigation of the mother's experiences. This study will focus on mother's experiences as they tend to be the main caregiver in families with a child with autism. Other possible mediating variables that will be investigated will include marital status, number of other children, age of child, age of child at diagnosis, length of time to get a diagnosis, satisfaction of services (formal support) received and satisfaction with informal social support.

The research objectives which will be tested by the quantitative questionnaires are listed below. The semi-structured qualitative interviews will follow up the questionnaires to explore what the findings of the quantitative data mean from a qualitative perspective, adding depth and richness to the statistical findings.

RESEARCH OBJECTIVES

1. To investigate whether mothers whose children they believe have high functioning autism or Asperger's syndrome but do not have a diagnosis will have higher self-reported stress than those whose children have a diagnosis of high functioning autism or Asperger's syndrome.

2. To investigate whether mothers whose children they believe have high functioning autism or Asperger's syndrome but do not have a diagnosis will have lower quality of life than those mothers whose children do have a diagnosis of high functioning autism or Asperger's syndrome.

METHODOLOGY

DESIGN

This cross-sectional study has used a mixed methods approach, run in two parts. Part 1 involved data collection via questionnaires which will mainly be subjected to one-way ANOVA's. Part 2 involved qualitative semi-structured interviews with a small sample of participants, data analysed using thematic analysis.

There was one <u>independent variable</u>: whether or not the mother has a diagnosis of high functioning autism or Asperger's syndrome for her child. There were 2 <u>dependent variables</u>: Mother's stress which has 3 sub-variables/levels; Parental Distress, Parent-Child Dysfunctional Interaction and Difficult Child and Health Related Quality of Life with 2 sub-variables/levels; Physical Component Summary (PCS-12) and the Mental Component Summary (MCS-12) score.

Other possible mediating *variables investigated included* marital status, number of other children, age of child, age of child at diagnosis, length of time to get a diagnosis/seeking a diagnosis, associated symptoms, satisfaction informal support received, satisfaction with formal social support.

PARTICIPANTS

The participants in this study were a convenience sample of mothers residing in the UK who have either received a diagnosis of high functioning autism or Asperger's syndrome in the last 5 years for their children or mothers who do not have a diagnosis for their children but a teaching, health or social services professional have suggested to them that their children may have high functioning autism or Asperger's syndrome and they are seeking a diagnosis. Men and other carers were not included in this study as it has been previously found that mothers are the ones who feel the impact of their child's autism (Gray, 2003). No incentive was offered for participation in this study.

Number of participants required: for the questionnaire part of the study, assuming that data was normally distributed, in order to detect a medium effect size (n = .15) for a between subject design to achieve power of .90 required 25 people in each condition (Clark-Carter, 2009).

Surveyed Participant's Characteristics: In total 84 participants completed the survey. Forty of whom were seeking a diagnosis for their child and 44 in the comparison group (those who had a diagnosis for their child). The average age of the mothers seeking a diagnosis was 35 and a half years of age (Std. Dev.: 5.38), and the average age of their child was 8 years of age (SD= 3.15). The average length of time of those seeking a diagnosis from when they first sought help to when they participated in this study was 4 and a half years (SD= 3.23). Fifty five per cent of the participants who were seeking a diagnosis for their child were working in either paid or voluntary employment and 85 per cent had a partner who was living with them.

The mean age of the mothers in the comparison group was 41 years (SD= 7.57) and the mean age of their children was 10 years (SD= 4.15). The mean age of diagnosis of the children of the comparison group was 7.9 years (SD= 3.2), and the average length of time taken to receive that diagnosis was 3.11 years (SD= 2.65). 54.5 per cent of those in the comparison group were working in either paid or voluntary employment and 75 per cent had a partner who was living with them.

Participant interviews: For the qualitative part of the study, 8 parents were interviewed, this was an opportunity sample, whereby all participants were given an opportunity to take part in the interviews and all of those who volunteered and met the inclusion criteria were interviewed. The inclusion criteria were women residing in the UK who were raising a child who had a diagnosis of autism or were raising a child for whom they were seeking a diagnosis of autism.   Four of these participants were seeking a diagnosis for her child at the time of the interview and 4 had already had a diagnosis for her child.

Interviewed Participant's Characteristics: The age range of the participants in the target group who were interviewed was 31 to 56 years, their child's ages ranged from 5 years to 14 years and time

taken to seek a diagnosis ranged from 3 to 12 years (7.25 years average), all the children were boys. Within this group three were living with a partner and only two were in either voluntary or paid employment.

The age range of the comparison group was 37 to 44 years, their child's ages ranged from 6 to 19 years of age, all the children were boys. The age at which the children were diagnosed ranged from 6 years to 10 years (7.75 years average). All participants in this group had a partner who was living with them and three were in either voluntary or paid employment.

MEASURES

The questionnaire pack consisted of 3 questionnaires: SF-12 Health Survey (Ware, Kosinski and Keller, 1996) the Parenting Stress Index Short Form (Abidin, 1995), and a demographic questionnaire which was disseminated via Qualtrics (see Appendix B).

12 Item Short-Form Health Survey (SF-12)

This is a 12 Item Short-Form measure of health related quality of life. The SF-12 takes approximately 3 minutes to complete and produces a Physical Component Summary (PCS-12) and a Mental Component Summary (MCS-12) score. When an individual respondent's scale score is below 45, or a group mean scale score is below 47 the health status is below the average range for the general population for each of the components (Ware and Sherbourne, 1992).

A high PSC-12 score would mean the participant would experience no physical limitations, disabilities, or decrements in their well-being, they would have high energy levels and their health would be rated as "excellent". A low PSC-12 score would mean the participant would have limitations in self-care, physical, social, and role activities, they would experience severe bodily pain, frequent tiredness and their health would be rated as "poor". The subscales measured physical functioning (i.e. ability to do all physical activities, including bathing or dressing), physical effects on

role (i.e. ability to work or carry out other daily activities as a result of physical health), bodily pain and general health.

A High MSC-12 score would mean an absence of psychological distress and limitations in their usual social/role activities due to emotional problems, and their health would be rated "excellent". A low MSC-12 score would mean the participant would suffer from frequent psychological distress, social and role disability due to emotional problems and their health would be rated "poor". The subscales measured vitality (energy), social function (i.e. ability or inability to carry out normal social activities due to physical and emotional problems), emotional effects on role (i.e. ability or inability to carry out work or other activities due to emotional problems) and mental health (i.e. any feelings of nervousness and depression)(Ware *et al,* 2002).

Test-retest reliability was established for the physical component with .89 and mental health component with correlations of .76 (n=232) (Ware, Kosinski and Keller, 1996) on a USA population. It was also compared against the SF-36 scale to establish concurrent validity in 14 tests which produced a median score of .67(Ware, Kosinski and Keller, 1996). This survey was further validated for item selection and scoring in 9 European countries including the UK (Gandek *et al,* 1998). Internal consistency was established with 3685 participants in Tehran with Cronbach's alpha's of 0.87 and 0.82 for the PCS-12 and the MCS-12 respectively (Asadi-Lari *et al,* 2010) and by the current study which found Cronbach's alpha's of .73 and .76 for the PCS-12 and MCS-12 respectively.

Although there is very little research in the area of health-related quality of life and parenting children with autism this survey has been used for parents raising school-age children with diagnoses of high functioning autism or Asperger's (Allik, Larsson and Smedje, 2006). It recorded a mean score of 44.7 for the PCS-12 and 49.1 MCS-12.

Parenting Stress Index Short Form (PSI/SF)

The PSI/SF (Abidin, 1995) consists of 36 items and produces scores for four subscales and yields from them a score of the overall level of total Stress that a person is experiencing as a parent. The subscales were as follows:

Parental distress (PD): This measure analyses feelings of parental incompetence, stresses associated with restrictions on lifestyle, conflicts with the child's other parent, lack of social support, and depression.

Parent-child dysfunctional interaction (P-CDI): This measure analyses a parent's perception that the child does not measure up to expectations and that interactions with the child are not reinforcing.

Difficult child (DC): This measure analyses behavioural characteristics of children that make them easy or difficult to manage.

Defensive responding (DR): Within defensive responding a low score will indicate that a parent is detached and uninvolved in parenting or that she is trying to present a favourable impression of herself.

The Parenting Stress Index has been well established as a measure for parenting children with Autism or Asperger's syndrome and mean total parenting stress scores range from 96.4 to 127 in these studies (Johnson *et al,* 2011, Epstein *et al,* 2008, Epstein *et al,* 2008, Bendixen *et al,* 2011, Bendixen *et al,* 2011, Braiden *et al,* 2012, Hoffman *et al,* 2009, Richardson, 2010, Hall and Graff, 2011a). The test retest reliability for this scale was assessed by its creators over a 6-month retest interval with N=270 and found reliability of .84 for Total Stress, .85 for Parental Distress, .68 for Parent-Child Dysfunctional Interaction and .78 for Difficult Child. The coefficient alpha was calculated on a sample size of 800 and reported reliabilities of .87 for Parental Distress,.80 for Parent-Child Dysfunctional Interaction, .85 for Difficult Child and the total stress was .91 (Abidin, 1995). Concurrent Validity was established against the long-form Parental Stress Index by Abidin (1995) with correlations of .82 for Parental Distress, .68 for Parent-Child Dysfunctional Interaction, .77 for Difficult Child and .94 for total stress, all of which are significant. Reliability was further

established in this current study and found coefficient alphas of .93 for Total Stress, .87 for Parental Distress, .84 for Parent-Child Dysfunctional Interaction and .86 for Difficult Child.

The Demographic Questionnaire

The demographic questionnaire (Appendix B), written by the researcher, consisted of questions concerning the participant's own personal circumstances and that of her child's diagnosis. Two further questions on the support received by the mother and how she rates her satisfaction with the support was included. These were extracted from the Emotional support sub-scale of the Perceived Support Scale (Krause and Markides, 1990). Internal consistency for this subscale was found with an internal consistency of alpha .925.

PROCEDURE

Ethical approval was applied for and gained from the Research Governance Board at the University of West of England (See Appendix E).

The researcher advertised for participants through online forums for parents of autistic children over a six-week period. Those mothers who were interested in learning more or interested in participating were directed to a participant information sheet on Qualtrics (Appendix A). They were then directed to an online questionnaire which included a participant information sheet, informed consent, a debrief statement and an invitation to volunteer to participate in the qualitative interviews (Appendix A and B).

Those who wished to participate in the interviews were invited to either leave their email address at the end of the survey or to email the researcher directly. When a participant indicated that they wished to participate in the interview the researcher contacted the participant via email initially to confirm their wish to participate followed by a phone call to arrange the time and circumstances of the interview. The interviews were conducted once analysis of the questionnaire was completed.

As participants resided in different areas of England and it was not always possible to conduct the interviews face-to-face, it was therefore decided that all interviews would be conducted over the telephone to ensure the conditions were the same for each participant.

INTERVIEWS

Following the questionnaires telephone interviews with 8 participants were carried out to further investigate the mother's stress and wellbeing considering their child's diagnosis or lack of diagnosis of autism or Asperger's syndrome. The purpose of these interviews were to further investigate the stresses and health related quality of life associated with diagnosis and parenting, if there were aspects of raising their child and the relationship to stress and quality of life that were not picked up on by the questionnaires and to provide further insight into these parents experiences.

In a systematic review by Novick (2008) it was found that data interpretation and quality of findings is similar in telephone interview to that of face-to-face interviews and telephones interviews have additional positive features such as decreased cost and travel, ability to reach geographically dispersed respondents, and enhanced interviewer safety.

In this study, to ensure there was no discrimination on the grounds of location, participants were geographically dispersed throughout the UK and were therefore interviewed via telephone for convenience and accessibility (see the interview schedule in Appendix C).

The questions were developed following analysis of the questionnaire data. This analysis found some significant differences between groups in some of the questionnaire data, therefore the interview questions were developed to explore these in more depth, particularly what the participants felt were the reasons between the differences. The questions were designed as open-ended to encourage rapport building between researcher and participant and to allow the participant to express their own perspective in their own words in as much detail as they wished (see Appendix G for a list of the interview questions). Each interview lasted between 20-45 minutes. These interviews were analysed using theoretical thematic analysis (Braun and Clarke, 2006).

Interviews as opposed to questionnaires and focus groups for the qualitative section of this research enabled the research to gain participant's views of their lives as portrayed by their own individual stories (Fossey *et al*, 2002). Semi-structured interviews were applied to allow for more focused exploration of the topic was required as a follow up to the specific issues that arose from the quantitative questionnaires.

ANALYSIS OF INTERVIEW DATA

Thematic analysis was the chosen method to organise and analyse the data. It was felt that this approach was best suited to this mixed methodology because of its flexible approach. Thematic analysis can provide a potentially rich, detailed and complex account of data, allow for any unanticipated insights within that data and enable the researcher to highlight any similarities or differences between the data sets (Braun and Clarke, 2006).

The 6 phases of thematic analysis (see appendix D) were followed: to ensure the researcher was very familiar with the interview data as required for thematic analysis the primary researcher conducted the interviews and transcribed them herself. The data was read and re-read before coding to ensure the researcher had immersed herself in the data. The data was reviewed at the semantic level for recurring themes that were related to the research questions and represented some level of patterned response or meaning (Braun and Clarke, 2006). Themes and subthemes are illustrated in Appendix F.

Theoretical thematic analysis was chosen to allow the researcher to conduct a more detailed analysis of the research questions, and provide depth to the questionnaire data. Reliability of the data was controlled for inter-rater reliability, whereby another researcher will also code the interview data independently.

Rationale for methods approach

Two methods of investigation were used to investigate the hypothesis, questionnaires and interviews. These were designed to be complementary, whereby both sets of data analysis will enhance the other, and will generate complementary insights to create a bigger picture.

The questionnaires were used to determine if those seeking a diagnosis do suffer higher stress and poorer quality of life, thus testing the magnitude of the hypotheses analysis with a representative sample. However these questionnaires were not able to investigate why this was the case and what the mother's experiences were. Therefore interviews were also conducted with a smaller sample of participants to allow a more in-depth exploration of their circumstances and their understanding of their situations.

Sampling Bias considerations

It is possible that the online sampling technique applied would be opened to sample selection bias. For example, because this research has recruited from online support forums, it is excluding those from the research population who do not use such forums. Those mothers may not use the forums because they do not know about them, they do not find them helpful or they do not like them, or they may not have the time or freedom to access them.

Self-selection bias may also apply as the research sample may have volunteered because they feel that the stress and quality of life that is being measured may correlate with how they are feeling and they wish to express that. This may especially be true of those who volunteered for the follow up interview because the interview would give them further opportunity to express their feelings about their levels of stress and quality of life in depth, in their own words. There may be mothers who are raising a child with autism (diagnosed and non-diagnosed) who cannot particularly relate to feelings of stress or poor health-related quality of life and therefore did wish to participate in an online survey and telephone interview.

ETHICAL CONSIDERATIONS

All information provided in the questionnaires was anonymous. The researcher had no way of identifying participants, unless they provided their email address at the end of the questionnaire to be contacted for an interview.

If at any point participants wished to withdraw from this study they were instructed to contact the researcher via email with the date and approximate time they filled in the survey and their information would be removed and deleted immediately. They were informed that they were not required to give a reason and their medical care or legal rights would not be affected (see Debrief Statement, Appendix A).

Any personal data obtained for the purposes of contacting the participants who volunteered for the interviews was kept separate from the transcripts of the interviews. All identifiable data was either not included or was coded where appropriate in the transcripts. Only the primary and secondary researchers have access to the interview data.

RESULTS

The results of this study are analysed and presented in two parts. The first part, the quantitative section, displays the data from the three questionnaires: the Demographic questionnaire, the Parenting Stress Index Short Form and the 12 Item Short-Form Health Survey. Comparison between the two participant groups were analysed using one-way ANOVAs and graphs used to visually illustrate any significant differences found.

The second part of this results section exhibits the qualitative input of this research, the analysis of the interview data. The interview analysis of the participant group who are seeking a diagnosis for their child is displayed first followed by the participant group who already have a diagnosis for their child. The information is presented under subsections which are the themes that emerged from the data.

Part 1: Quantitative Results

The differences between the group of mothers who did not have a diagnosis for their child and those who did have a diagnosis for their child was analysed using one-way ANOVAs. The results are listed below.

THE DEMOGRAPHIC QUESTIONNIARE

The Demographic questionnaire was studied for differences between groups which would highlight any confounding factors.

Mother's age

The average age of the group of Mothers who were seeking a diagnosis was 36 years old (SD= 5.38), 41 years old for Mothers who had a diagnosis for their child (SD= 7.57). There was a significant difference between these groups at $p<.05$ level for the two conditions [$F_{(1, 82)} = 11.25$, $p = 0.001$]., those mothers with a diagnosis being significantly older.

Child's age

The average age of the group of children who were seeking a diagnosis was 7.8 (SD= 3.15) and 10.8 for children who have a diagnosis (SD= 4.15). There was a significant effect between these groups at $p<.05$ level for the two conditions [$F_{(1, 82)} = 9.79$, $p = 0.002$], those children with a diagnosis being significantly older.

Number of other children

The number of other children the Mothers were raising was also recorded. The mean number of other children for the group of mothers who were seeking a diagnosis for their child was 1.5 (SD= 0.49). The group of mothers who had a diagnosis for their child had a mean of 1.7 other children (SD= 0.48). The ages of the siblings ranged from under 1 year old and over 18 years in both groups.

Working Status

Mothers were asked if they were employed, either paid or voluntary. 22 (55%) of the mums who were seeking a diagnosis stated that they were employed, 24 (54.5%) of the mothers who had a diagnosis stated that they were employed.

Marital Status

When asked for their marital status, 34 (85%) of the mums who were seeking a diagnosis stated that they had a partner who was living with them, 33 (75%) of the mothers who had a diagnosis stated that they had a partner who lived with them.

Age of child when parents first noticed symptoms

Mothers were asked how old their child was when they first noticed something was wrong. The mean age of the child who did not have a diagnosis was 2.35 (SD= 2.68) and 2.43 for those who received a diagnosis (SD= 1.79). There was not a significant difference between the groups at $p<.05$ level [$F_{(1, 82)} = 0.027$, $p = 0.87$].

Age of child when parents first sought help

Mothers were also asked how old their child was when they first sought help. The mean age of the child who did not have a diagnosis was 3.45 (SD= 3.02) and 3.41 for those who received a diagnosis (SD= 2.69). This again was not a significant difference between the groups at p<.05 level [$F_{(1, 82)}$ = 0.004, p = 0.948].

Satisfaction of social support received

Mothers were asked to rate their satisfaction with the informal social support, and emotional/tangible support they received in a scale of 1-10 where 1 was very dissatisfied and 10 was very satisfied. The mean rating from mothers who did not have a diagnosis for their child was 4.10 (SD= 2.95) and for those who did have a diagnosis for their child was 4.16 (SD= 2.66). This difference was not found to be significant at p<.05 level [$F_{(1, 81)}$ = 0.110, p = 0.741]. This suggests that there was no statistically significant difference between the groups in their satisfaction with the informal support and emotion/tangible support they received from friends and family members.

Mothers were then asked to rate their satisfaction with the formal support they received. The mean rating from mothers who did not have a diagnosis for their child was 4.28 (SD= 3.12) and for those who did have a diagnosis for their child was 4.07 (SD= 3.10). This difference was not found to be significant at p<.05 level [$F_{(1, 81)}$ = 0.008, p = 0.927].

Socioeconomic Status

The socioeconomic status of both groups of mothers was not measured in this study, therefore no comparison was made between the groups.

PARENTING STRESS INDEX SHORT FORM (PSI/SF)

This questionnaire had 4 sub-scales: Parental Distress, Parent-Child Dysfunctional Interaction, Difficult Child and Defensive Responding. A total parental stress score was also measured.

Parental Distress

The first subscale measured in the Parenting Stress Index between the groups was Parental Distress. The mean score for the experimental group was 43.93 (SD= 8.42) and 39.34 for the comparison group (SD= 9.46). According to Abidin (1995) parents who obtain a score of 33 or more are experiencing clinically significant parental distress meaning that they are highly distressed by their functioning in the parental role. This was 98% of participants who are seeking a diagnosis and 80% of participants who have a diagnosis for their child.

Using a one-way ANOVA the difference was found to be significant between these groups at $p<.05$ level [$F_{(1, 82)} = 5.458$, $p = 0.022$]. This means that the group of mothers who were seeking a diagnosis had significantly higher levels of feelings of parental incompetence, stresses associated with restrictions on lifestyle, conflicts with the child's other parent, lack of social support, and depression than the group of mothers who already had a diagnosis for their child.

The frequency of these scores and their normal distribution curves are displayed in histograms in figures 1 and 2.

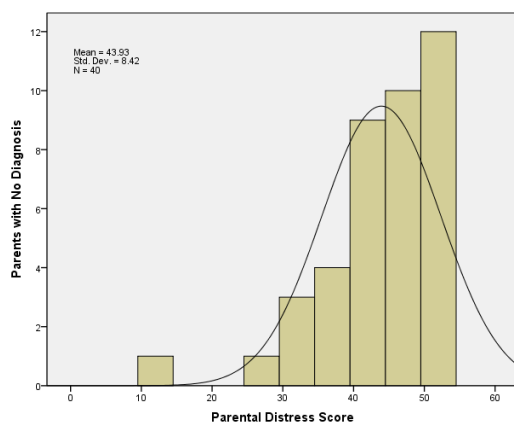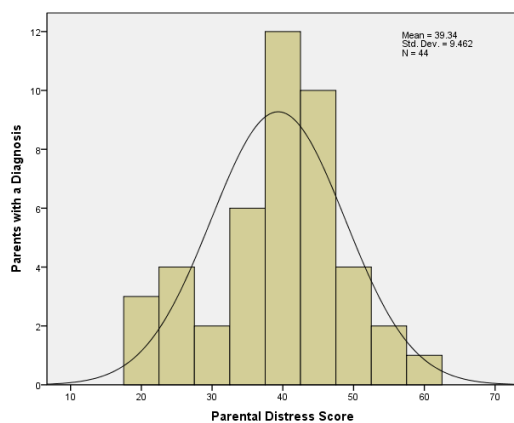**Figure 1***: Parenting Stress Index Parental Distress Scores for Parents with No Diagnosis (n= 40)*

Figure 2*: Parenting Stress Index Parental Distress Scores for Parents with a Diagnosis (n= 44)*

Parent-Child Dysfunctional Interaction

The mean score for the P-CDI for the experimental group was 37.13 (SD= 8.93) and 33.25 for the comparison group (SD= 9.20). A score of more than 26 is clinically significant meaning that the mothers are highly distressed by the quality of their parent-child interaction (Abidin, 1995). Ninety eight per cent mothers who are seeking a diagnosis had a score of 26 or more, and 82% of the comparison group scored 26 or more.

The differences between the groups were analysed using one-tailed ANOVA and were found to be closely approaching significance [F(1, 82) = 3.823, p = 0.054].

Difficult Child

The Difficult Child mean scores for the group of mothers seeking a diagnosis was 46.77 (SD= 7.73), and 42.86 for the comparison group (SD= 8.02). All participants seeking a diagnosis and 89% of the comparison group had a score of more than 33, which indicates a clinically significant score (Abidin, 1995). A clinically significant score means that the parent sees her child as possessing many disruptive behavioural characteristics (Abidin, 1995).

The differences between the groups were analysed using one-tailed ANOVA and were found to be significant at p<.05 level [F(1, 82) = 5.158, p = 0.026], therefore those who were seeking a diagnosis for their child rated the behavioural characteristics of their child as significantly more difficult to manage than those who already had a diagnosis for their child.

The frequency of these scores and their normal distribution curves are displayed in histograms in figures 3 & 4.

Figure 3*: Parenting Stress Index Difficult Child Scores for Parents with No Diagnosis (n= 40)*

Figure 4*: Parenting Stress Index Difficult Child Scores for Parents with a Diagnosis (n= 44)*
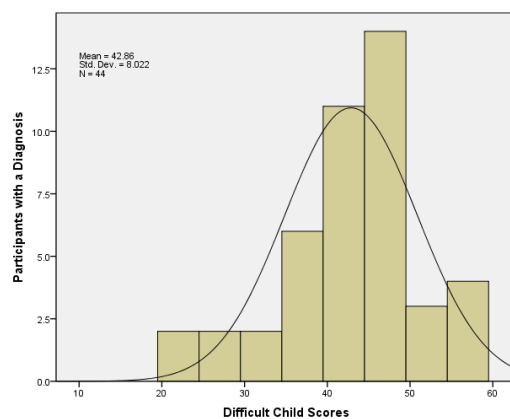
Overall Stress

For both groups the overall parental stress was very high. The mean total score for the experimental group was 127.83 with a standard deviation of 21.26. The highest possible score is 180 and the lowest is 0. According to Abidin (1995) parents who obtain a total score of 90 or more are experiencing clinically significant levels of stress. This was true of 95% of the participants seeking a diagnosis for their child. Other studies which measured Total PSI of parents raising children with Autism found similar results with 75.7%, 96.4% and 80.6% of participants scoring clinically high stress levels (Epstein *et al,* 2008, Braiden *et al,* 2012, Braiden *et al,* 2012, Richardson, 2010).

The mean total score for the comparison group was also very high with 115.45 and a standard deviation of 23.20. Eighty four per cent of this group had a score of 90 or above thus exhibiting clinically significant levels of stress (Abidin, 1995).

Using a one-way ANOVA the difference was found to be significant between these groups at p<.05 level [$F_{(1, 82)}$ = 6.448, p = 0.013], this meant that the group of mothers who were seeking a diagnosis reported significantly higher stress than the group of mothers who already had a diagnosis for their child.

The frequency of the scores and the normal distribution curve are displayed in histograms in figures 5 and 6.

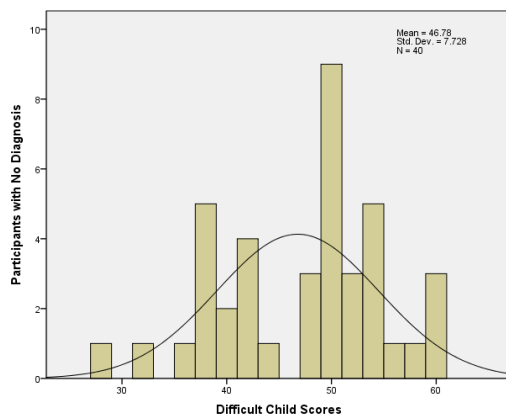**Figure 5***: Parenting Stress Index Total Scores for Parents with No Diagnosis (n= 40)*
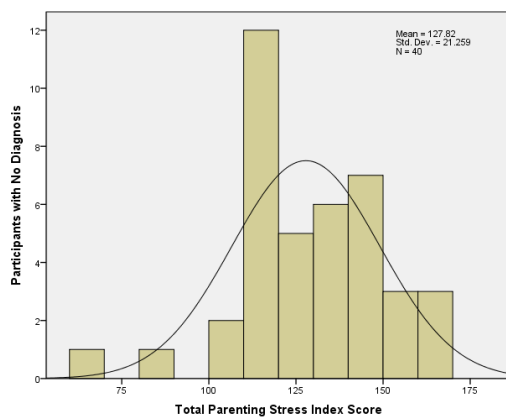
Figure 6*: Parenting Stress Index Difficult Total Scores for Parents with a Diagnosis (n= 44)*
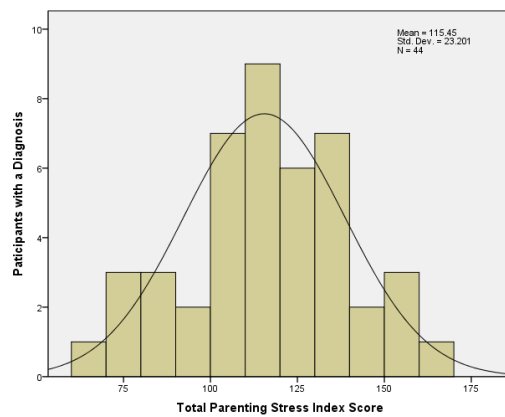
Defensive Responding

No participants scored less than 10 in the Defensive Responding category of the PSI/SF, demonstrating that all participants answered truthfully.

Parenting Stress Index Summary

The summaries of the mean Parenting Stress Index Short Form (PSI/SF) scores are tabulated in Table

1.

Table 1

Summaries of the mean Parenting Stress Index Short Form scores

	Participant Group	Mean	Standard Deviation	Score Indicating Clinical Significance	% Clinically significant	*P* value
Parental Distress	*No Diagnosis*	43.93	8.42	>33	98%	0.022*
	Diagnosis	39.34	9.46		80%	
Parental-Child Dysfunctional Interaction	*No Diagnosis*	37.13	8.93	>26	98%	0.054
	Diagnosis	33.25	9.20		82%	
Difficult Child	*No Diagnosis*	46.77	7.73	>33	100%	0.026*
	Diagnosis	42.86	8.02		89%	
Overall Stress	*No Diagnosis*	127.83	21.26	>90	95%	0.013**
	Diagnosis	115.45	23.20		84%	

*p<0.05, **p<0.01

Relationship between mother's age, child's age and the Parenting Stress Index scores

There was a significant difference between the groups in mother's age and the child's age at data collection. Therefore to gain an estimate of the degree of association between these demographic factors and the Parenting Stress Index scores a correlation for each subscale was conducted.

For the mother's age category no correlation was found for Parental Distress [r = -.176, n = 84, p = .109], Parent-Child Dysfunctional Interaction [r = -.069, n = 84, p = .532] or Total Parenting Stress scores [r = -.189, n = 84, p = .86].

For the child's age category no correlation was found for Parental Distress [r = -.172, n = 84, p = .118], Parent-Child Dysfunctional Interaction [r = .118, n = 84, p = .287], Difficult Child [r = -.112, n = 84, p = .310] or Total Parenting Stress scores [r = -.061, n = 84, p = .581]. However, the Difficult Child subscale was found to be significantly correlated with the mother's age [r = -.257, n = 84, p = .018]. This has been demonstrated in a scatter graph below (Figure 7).

Figure 7*: Correlation of the Parenting Stress Index Difficult Child Scores (n= 84)*
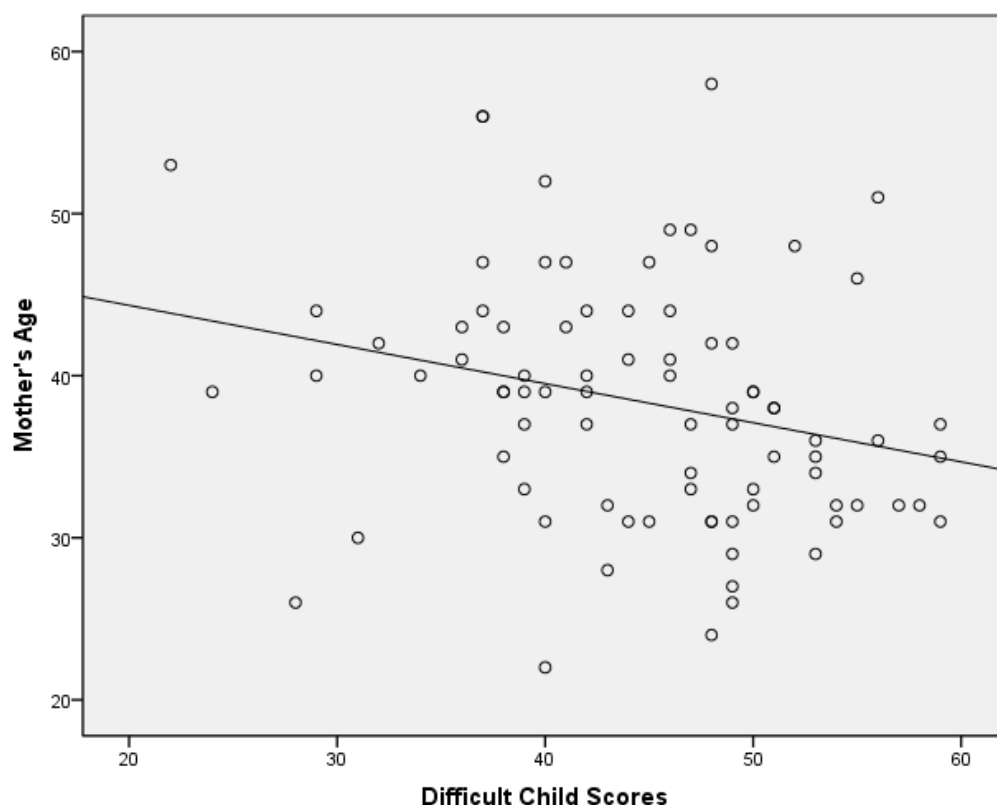
12 ITEM SHORT-FORM HEALTH SURVEY (SF-12)

Parents were also asked to fill in the SF-12 to measure their health-related quality of life. This scale measures the physical (physical component summary) and the mental health (mental component summary) components of quality of life. Interpretation of this survey uses norm-based scoring whereby each scale is scored to have the same average (50) and the same standard deviation (10). Whenever an individual respondent's scale score is below 45, or a group mean scale score is below 47, health status is below the average range for the general population for each of the components (Ware and Sherbourne, 1992).

Physical Component Summary (PCS)

The mean score for the PCS for the experimental group was 49.95 (SD= 13.81) and 46.07 for the comparison group (SD= 11.87). Therefore only the comparison group was found to be below the average range for the general population, meaning that they would have limitations in self-care, physical, social, and role activities, they would experience severe bodily pain, frequent tiredness and their health would be rated as "poor".

The differences between the groups were analysed using one-tailed ANOVA and were found not to be significant at $p<.05$ level [$F(1, 82) = 1.915$, $p = 0.170$].

Mental Component Summary (MCS)

The mean score for the group of mums seeking a diagnosis for their child was 29.58 (SD= 10.98) and the mean score for the comparison group was 38.81 (SD= 10.91). Therefore both groups indicated much lower mental component summary mean scores than the general population. This result contradicts the result from the study by Allik *et al* (2006) which found that parents raising a child with high functioning autism scored much higher with 49.1 on the MCS-12.

The differences between the experimental and comparison group were analysed using one-tailed ANOVA and were found to be significant at p<.05 level [F(1, 82) = 15.057, p = 0.000]. This means that the group of mothers who were seeking a diagnosis for their child scored significantly lower in the mental component summary than those with a diagnosis, therefore they are reporting more frequent psychological distress, and more social and role disability due to emotional problems (Ware *et al,* 2002).

The frequency of the MCS scores and the normal distribution curve are displayed in the histograms below (figures 10 and 11).

Figure 8: *SF-12 Mental Component Summary Scores for Parents with No Diagnosis (n= 40)*
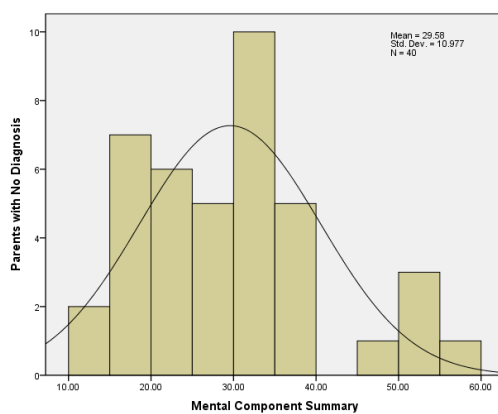
Figure 9: *SF-12 Mental Component Summary Scores for Parents with a Diagnosis (n= 44)*
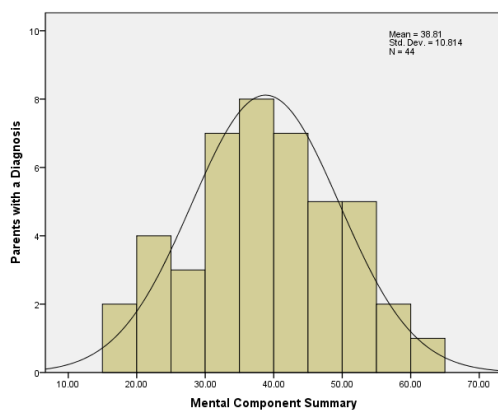
Health Survey Scores Summary

The summaries of the mean 12 Item Short-Form Health Survey (SF-12) scores are tabulated in table2.

Table 2:						
Summaries of the mean 12 Item Short-Form Health Survey scores						
	Participant Group	**Mean**	**Standard Deviation**	**Individual score indicating below average range for general population**	**% Below average range for general population**	***P* value**
Physical Health Summary	***No Diagnosis***	49.95	13.81	<45	30%	0.170
	Diagnosis	46.07	11.87		86%	
Mental Health Summary	***No Diagnosis***	29.56	10.98	<45	70%	0.000**
	Diagnosis	38.81	10.81		39%	

*p<0.05, **p<0.01

Correlations between mother's age, child's age and the Health Survey scores

To get an estimate of the degree of association between demographic factors and the Health Survey scores a correlation for each component was conducted. For the mother's age category no correlation was found for Physical Health Summary scores [r =.102, n = 84, p = .356] or the Mental Health Summary Scores [r = -.191, n = 84, p = .082]. For the child's age category again no correlation was found for Physical Health Summary scores [r =.012, n = 84, p = .911] or the Mental Health Summary Scores [r = -.114, n = 84, p = .301].

Therefore the mother's age and child's age factors were not significantly related to the comparison of the Health Survey component scores between the groups.

Part 2: Qualitative Results

PARENTS WHO ARE SEEKING A DIAGNOSIS FOR THEIR CHILD

In all, 4 parents in this group were interviewed; their details are presented in Table 3, however socioeconomic status not measured. Following the thematic approach of analysis the researcher became familiar with the data by transcribing and reading and re-reading the interviews. The data was then coded and collated into relevant themes. Four key themes emerged in the analysis namely *beliefs that their child has autism, interactions with teaching and health professionals, expectations on what will change with a diagnosis, health and quality of life*.

The themes illustrated ways in which the difficulty of getting a diagnosis and the difficulties they are encountering when raising their children and needing help had affected their stress levels, mental and physical health and their quality of life. Some of these themes will be further divided into sub-categories.

Table 3:

Details of interviewed participants seeking a diagnosis for their child

	Demographic information					Health Survey		Parenting Stress Index			
Participant	Age	Child's age	Time spent seeking a diagnosis	Employment Status	Living with a partner?	MCS	PCS	PD	P-CDI	DC	Total PSI
A	56	14	8	Employed	Yes	39.99	40.05	43	29	42	114
B	31	5	3	Unemployed	No	23.41	25.23	45	35	49	129
C	36	12	7	Unemployed	Yes	28.88	31.44	48	48	53	149
D	42	10	7	Employed	Yes	35.28	32.82	44	34	51	129

BELIEFS THAT THEIR CHILD HAS AUTISM

All the participants describe difficulties their children are having which they attribute to ASD. They each describe their children's difficulty with developing their speech as one of the first indications of a problem: "*He worked very hard at home; there was talk of sending him to a specialised language unit because he was so severely behind but obviously very bright*." (A) Three of the participants' children had actually been referred to and attended speech and language therapy sessions. They all talked about aspects of their child's behaviour that was unusual when compared to other children they knew, for example aggression was a factor described by three participants:

"*Things were getting so bad at home, it was horrific. Every weekend he'd wake up at 3am, literally tearing his room apart. Shouting, biting, screaming, physical violence*." (B)

"*For example, he got into trouble at school… he was punching, biting, scratching, kicking. That night I eventually got him to sleep at 2am and he was up again at 3:20am. I didn't do it, I didn't do it.*" (D)

Other traits mentioned by the participants include sensory problems, sleeping problems, hand flapping and little eye contact and problems making friends and fitting in at school. Two examples include:

"*We first noticed problems with our son when he was about 18 months old. Em… he wasn't like any other child, he was really aggressive. He didn't like hugging. He stopped walking, talking, he would eat things that he shouldn't be eating. Things like wood.*" (B)

"*It was year after year of him not fitting in, he would get over excited when he saw another child and jump up and down and flap his hands. The other child would then think that I am going to avoid that child, that is a bit weird. We have had to teach him how to interact with people.*" (A)

All mothers discussed noticing that their child was not developing as other children were:

"I thought I was the most brilliant mother because I had this baby who would sleep all day and he would stare up into the lights quite contentedly, quietly… but other people were seeing different things because they had different experience than me and they saw this child who was staring up at lights and realised he wasn't quite developing in the normal way." (D)

"It is just the entire level of emotional support that he desperately needs. I noticed that with other children you don't necessarily have to teach them the same things." (C)

There was a certainty from all of the mothers that their children did have some form of high functioning autism or Asperger's syndrome and they did not seem to be looking for the diagnosis for confirmation, more so that they could get recognition from professionals who could help them, schools in particular. There was a sense that they had already accepted that their child did have HFA/Asperger's and that they were bewildered by the fact that this is not being recognised by professionals. Only one of the parents mentioned any doubt in her belief that their child had ASD:

"The thing is, even I doubt sometimes, I look at him when he has had a good day and think he can be any child and I look at him when we try and get out to a supermarket and he is falling apart in the shop and saying it's too loud, and he's ripping his ear defenders off and that kind of thing then I think well maybe not. And I think its natural but in the main we don't have any doubts. I think its other people that doubt us a little bit." (D)

Because of this certainty three of the mothers interviewed were finding it useful to treat their child as if they have ASD even though they did not yet have an official diagnosis. This was helping them manage their child's behaviour and seemed to give them something to focus hope on. One parent was even advised by their son's educational psychologist. "*I did ABA therapy with autism and I was doing that at home. He was ticking many of the boxes for high functioning autism or Asperger's, I*

could see that so I was going to treat it like that. Let a professional tell me it's not and I could see

that it is. Something has got to be able to help him." (B)

INTERACTIONS WITH TEACHING AND HEALTH PROFESSIONALS

Frustrations in interactions with teaching and health professionals

The interviews revealed that the all the mothers had a strong belief that their child in fact did have autism and these beliefs have either been instigated or strengthened by a teaching or health professional at some point in their child's development, for example:

"*The educational psychologist was very encouraging; she virtually said to us that we should treat him as having Asperger's*" (B)

"*his reception teacher pulled me aside and said I want you to know that he reminds me of children I have taught with Asperger's, you might want to get that checked out*." (A)

It became apparent that when seeking a referral for assessment of ASD for their child these parents have interacted with a wide range of health professionals, these include medical professionals such as health visitors and general practitioners, allied health professionals such as speech and language therapists, teaching professionals and psychologists. Although some of these interactions have been quite helpful in taking a step towards the ASD assessment the participants mostly discussed frustrations and slow progressions in their interactions.

"*The educational psychologist said there was no issues, the Health Visitor completely ignored me, the speech and language therapist said verbal dyspraxia, the paediatrician wouldn't accept it at first, made us wait 6 months and we are currently waiting to see if CAMHS will accept our referral.*" (B)

All participants describe instances whereby their concerns have been dismissed by professionals they are seeking help from with comments such as "it's the terrible twos" and "he's a dreamer". They discussed meetings with professionals who fail to see the problems the participants see: *"[The community paediatrician] looked at him briefly and he said he is definitely not autistic because he*

gave me a second of eye contact." (C) There was a sense from all the mothers that they have felt powerless to argue with the opinions of the health professionals even though they are very confident in their belief of their child's condition and their ability to see the autistic traits that the professionals are not seeing.

Two of the participants described interactions with teaching professionals that they found particularly frustrating and upsetting:

"In reception we asked if he was ok because we knew he wasn't, and the teacher said he was lovely and that was it. We had learned from pre-school, they hadn't told us that he was lying on the floor at times, not being involved or responsive or useful at all. It would have been handy if they had told me that! In year 2 the teacher basically called him a chore and we had been pulled over the coals as parents because they said his discipline was terrible." (C)

"I still couldn't believe they couldn't see any of this at school. They had labelled me as neurotic and kept telling me he was fine." (B)

Three parents have been referred to (and two have attended) parenting classes to address their child's behaviours by teaching and health professionals. Those parenting classes have been described by the interviewee's as useful in that they helped confirm to the participants that they were doing the right things with regards to addressing their children's behaviours such as discipline techniques. In two of the cases it has further confirmed to them that their child's behaviour is not quite right. But overall they have felt these classes were not appropriate for them as they did not teach techniques to deal with children displaying the autistic traits they believed their children had.

EXPECTATIONS ON WHAT WILL CHANGE WITH A DIAGNOSIS

All participants discuss a sense of relief if they receive a diagnosis, as they will have an explanation for their child's struggles and behaviours which they can use when they feel they are being judged as parents: "*when people are staring at you in the street or judging you, I can say that this is autism*" (C). Three of the mothers talked about how they hoped that a diagnosis will bring about more

support from the schools although they all seemed unclear as to what they can expect outside of the schools:

"I think there will be more support available; it will take the pressure off from school. When we have a formal diagnosis that is NHS approved and stamped and can't be ignored, I can legally do something about it if they are not following his statement. I will be able to help with things like DLA." (B)

"I hope we get some support, but I don't imagine a diagnosis will change much and we will have to fight for any help we get. There is a common perception among parents that after your diagnosis the attitude is 'don't let the door hit you on the way out'." (D)

Two of the mums hoped that a diagnosis would help them meet other parents with children with autism: *"there are some programmes for ASD that only accept kids with a diagnosis, it would be nice to meet like-minded families we can just chill out with"* (A).

PHYSICAL AND MENTAL HEALTH

Stress

With all of the participants there was a sense that raising their child whom they believe has autism has had an impact on their mental health. The stress involved in raising their child was discussed and the worry for their child at school and for their future was a major cause of stress for them as was the feeling of helplessness as they describe not knowing how to help their child and how to make things better for them:

"It is very stressful because you also don't have any support or any help about how you deal with your child and how you make things better. He is quite a miserable child because the world is a scary place for him and without any outside help you don't have any expert to say this is how you can make his life a bit happier." (A)

"You don't have that relaxed now he is in somebody else's care feeling. You worry all the time, day and night." (D)

The child's behaviours and how they have impacted on their everyday lives was a topic of discussion for all of the mothers as they felt they were unable to lead what they thought were normal lives, for example, simple daily chores and errands would have to follow a routine and their mentions of the inability to do spontaneous things: *"Even ordinary things that people take for granted, need military planning. It is very stressful.*" (C)

All participants seeking a diagnosis talked about the general on-going stressors they are experiencing raising their child who they believe have ASD, most particularly an inability to connect with their child and help them or calm them down when they are stressed or having a tantrum:

"At one point for 3 months he was only sleeping up to 2 hours a night. Some of it is because when he has bad days they can be really really bad…we don't even know what started it off. It could be from 2-3 days ago. And having 2 children at the same time, the stress of the guilt, he takes a lot of my time and energy from my other children" (B)

"The most stressful is the when we want to talk to him, he just shouts and threats. You can see in his head that he just can't cope with the ideas that." (D)

All of the mothers also described the problems they have experienced in seeking a diagnosis as a stressor. They all talked about how they struggled to cope with their child's behaviours and worried for their child, and the fact that they were not getting recognition for what they are going through further impacted on their feelings of stress:

"It is the fact that you feel you are banging your head against a brick wall. You feel you are getting nowhere. You are getting somewhere but you do not see it." (B)

"Families that are already under stress because they have got a heavy parenting load, it is too much. I don't want to take [seeking a diagnosis] all back up again, the last time I felt my chest tightening, chest pains. I took a break from work." (A)

They talk about how not having an explanation for their child's behaviour has also been stressful as it has led to judgements on them as parents when their children don't behave as expected in public *"…it is stressful not knowing and not having a diagnosis because you can't then explain to people what is going on. You can't say to people 'oh he is behaving like this because of this', you haven't got piece of paper that backs you up…"* (D)

Depression

Depression was discussed by all participants, all of whom are taking medication to manage their depression. All mums seeking a diagnosis described how the stress of raising their child has led to their depression: *"…I definitely feel myself sinking into depression, without a doubt. It was a case of I don't care anymore. I had to keep reminding myself that it's not my fault. The lack of sleep, it got to the point that I was hallucinating, he was sleeping less than 2 hours a night and that wasn't even in one stretch…. I would definitely say that it was stress to the point of depression, I could definitely see the dark side.*" (C)

All four mothers attributed the process of trying to get a diagnosis as adding to their depression. They discussed the frustrations and stress they experienced when they felt they were being dismissed by the professionals they spoke to or the fact that they felt they were blamed for their child's behaviour and how this was leading to their depression: *"It can get me down because you don't feel like you are being listened to. Your self-esteem starts to go down. You start to doubt what is normal and what isn't. You can't trust your own judgement when you go get help."* (D)

When discussing depression two mothers talked about how interacting with their child was not as they had expected and they were not able to engage with their child as they felt they should:

"*I can't engage with my son the way I envisaged. I always have to be calm. I feel a lot of times I am just trying to manage him throughout the day. I can't just change his routine, I have to be careful. You have to calculate your every move with him, you can't let him get too excited, it gets way too much!*" (C)

"*It was almost as if it was really hard to love him because he gave me so little back. He never had been very affectionate me.*" (D)

QUALITY OF LIFE

The participants also mentioned how raising their child has impacted on other areas of their lives. Isolation was a big factor in these conversations from all of the parents. This was because they felt their child was finding social situations stressful, they were not fitting in as a family at social events and they stopped inviting others to their home as they felt their friends did not understand their child's behaviours and needs:

"*If there was a family party, he wouldn't go. On occasion, there was a christening. He stood with his back against the wall, swaying from side to side, saying 'are we going now, are we going now'. And this carried on until we left. The same happened at wedding function we went to.*" (B)

"*We don't ask anyone over. We ask friends who haven't any children but they haven't got children with autism, and they don't understand. If you don't have a child like that then it is very hard to understand one.*" (C)

All mothers seeking a diagnosis also talked about how they feel they are not able to leave their child in the care of others (when they are not at school) and this has impacted on their work choices and their ability to enjoy a social life away from their child. There seemed to be a strong feeling of

unending sacrifice from the mothers in this group for their children which was making them unhappy:

"I had to give up my job and work very part-time from home. This is because I was having to leave work to pick him up from school early if he soiled himself or had to take him to any medical appointments" (D)

"I don't work, I look after him. When he was younger, I used to study at the OU, but he got so hard now that I don't got no time at all now so I had to give that up at the end of the second year. I kept hoping that the time will come where I can go back but I don't see that happening any time soon." (C)

"We can't get a normal baby sitter in. In all honestly some of it is probably me, there are probably people out there who could be brilliant with [my son] but I have very high standards and I would worry about it and not enjoy the time out anyway." (D)

"I think I have gone out about 4 or 5 times in [my son's] life. I did go away on holiday for a day and a half and got called back. But other than that I have just been with him." (B)

PARENTS WHO HAVE A DIAGNOSIS FOR THEIR CHILD

Four parents in the participant group who had a diagnosis for their child were interviewed, their details are presented in Table 4, however socioeconomic status was not included. Three key themes emerged in the analysis, some of which were similar to the participant group who were seeking a diagnosis for their child, these were: *struggles before the diagnosis, diagnosis, health and quality of life*.

The themes showed ways in which the difficulty of getting a help for their child prior to diagnosis and receipt of diagnosis has impacted on their lives and how raising their child has impacted on their mental and physical health and quality of life.

Table 4:

Details of interviewed participants who have diagnosis for their child

	Demographic information					Health Survey		Parenting Stress Index			
Participant	**Age**	**Child's age**	**Age of child at diagnosis**	**Employment Status**	**Living with a partner?**	**MCS**	**PCS**	**PD**	**P-CDI**	**DC**	**Total PSI**
E	39	13	10	Employed	Yes	51.42	55.90	34	27	40	101
F	42	12	7	Employed	Yes	45.09	46.20	24	26	37	87
G	44	19	8	Employed	Yes	47.41	51.75	41	34	44	119
H	37	6	6	Unemployed	Yes	56.80	35.35	22	22	29	73

STRUGGLES BEFORE THE DIAGNOSIS

Noticing their child had problems

All of the mothers interviewed noticed their children were having problems and were seeking help and advice for this, they noticed behaviours before their children were of school age which they described as unmanageable, difficult or unusual. The behaviours ranged from aggression, head banging, biting, sensory problems and social problems: "*When he got to 18 months old he would scream and cry and head bang and do all these really distressing things to watch. He was biting himself. I just wanted my child back and I don't know what had happened. At first we thought it was the terrible twos, but it never went away" (G)*. They discussed finding children increasingly upset or anxious as they got older, particularly in public or social situations:

"*Then we'd go to somewhere like the supermarket and there would be an absolute melt down and he would be 7, 8 ,9 years old and he'd be having this massive tantrum in the supermarket." (E)*

"*He was very sociable and very much liked, but he had trouble going to people's houses and staying overnight. Although he wanted to, he used to tell us that he never slept. At the time didn't really believe him, but I think now he didn't… in hindsight.*" *(F)*

Problems with school

All mothers in this interview group discussed problems becoming increasingly noticeable and unmanageable when their child attended school:

"*It wasn't until he started school, it wasn't too bad in reception but when he went into year 1 he entered the more formal setting. He became very unhappy and started to show signs of very difficult behaviour we had never seen from him. He was 5 years old, only just 5." (F)*

"*Year 1 was a complete nightmare, probably one of the worst times in probably both of our lives and he had started seeing the SENCO, and he was having to be kept inside during play time because he was hurting some people.*" *(E)*

"His first day back at school was always bad and at the end of the week he would end up not speaking to us, and we wouldn't be speaking to each other. There was so much stress and he couldn't do anything, everybody would be in tears apart from my son." (G)

"As soon as he started school he became horrendous at home. I made the decision he wasn't coping and so I left my job so I could drop him off and pick him up from school so he could be with me more. I felt like I was giving it up for him but in hindsight I have made the right decision." (H)

Some talked about how their children were unhappy at school: *"My son didn't act out of school, so he was fine at school, but then when he got home he would be crying and saying I hate school and everything else"* (G). Two of the mothers discussed frustrations on how their children were getting into trouble at school for naughty behaviour or not keeping up with school work when they felt it was clear that their child was having problems:

"School now acknowledge that he is different, but you know he was completely used to being told off but for bad behaviour that he became completely desensitised to the telling off." (E)

"We discovered from other children that he was being shouted at and he was being put in a cloakroom and left there to do his work. Because he had mental problems he was very slow to write and we couldn't figure out what was wrong. We assumed it was a clash with the teachers." (F)

Three of the mothers described having difficulties getting understanding from the teachers at their child's school. They had felt they were being treated as they were overreacting or oversensitive to their child's inability to cope at school and there was a sense that they did not feel they were being taken seriously when they raised their concerns: *"Before the diagnosis I we just don't feel that the school have taken us seriously all the way through… it will be good to be able to come into the school with that piece of paper that says something, that will include the handwriting problems and all…we are hoping that the school are going to nurture him a bit more now."* (H)

Two of the mothers felt they were being blamed for their child's behaviour prior to receiving the diagnosis:

"School were basically persecuting me constantly, because he was hitting other people and cursing. School were at that point were talking to me like I was causing his violence." (E)

"In Year 1, when his behaviour got even worse and I was told to get my child to the doctor and sort his behaviour out" (F)

Trying to get recognition of child's problems

All participants described a long time between realising their child was having difficulties to receiving a diagnosis for their child but they do not discuss this in much detail. Those who realised their children had autism before the actual diagnosis realised this only a short time before. Therefore these mothers did not discuss stress of trying to get recognition of the ASD, but they did discuss difficulties in trying to get recognition that there was a problem and learning how to help their child, although again this is not described in much detail by the participants:

"We had gone through this cycle that I would feel that I had gone to the doctors and, it could be just my paranoia but, they'd roll their eyes and go 'oh no, not again'. And honestly he was banging his head repeatedly on the side of his cot and it was really distressing." (H)

"Actually getting a diagnosis is the hardest and most stressful part and until you do get that diagnosis you get an awful lot of… em… rolled eyes sort of feel, it's sort of 'aw it's that woman again and she is moaning that her sons got this and what's she going on about' sort of thing." (F)

Two mothers discussed the topic the diagnosis as a label and how that label was a positive thing in helping them to move forward in coping with their child:

"I hate this because one thing I keep getting from professionals is 'why do you want to label them.' I hate it. One of the things I would say to them is that you wouldn't give epilepsy drugs to a diabetic

because they are diabetic, you would find out what it was so you can treat it. I don't get this concept of let's not give them a label because you can't put things in place to help them. It is very difficult to get any of the services without the label." (H)

"The diagnosis helps in that giving it a name that you can say to people 'look, he isn't just a teenager, he isn't just lazy he has Asperger's, and if I can't actually describe that to you you best go and look that up.'" (E)

DIAGNOSIS

Receiving the diagnosis

All the mothers describe receiving the diagnosis as a very positive step forward and a relief: "*Honest to god I don't remember hearing anything else because I was quite emotional and crying, it was like a wave of relief. I don't know what it means. I am just relieved now that school now have to listen to us. That will be the biggest thing.*" (H)

They each describe the diagnosis as a positive life changing event as it gave them and others understanding of their child's behaviours and struggles:

"In a way it made things better, again things did get worse with my son, but this time [the diagnosis] gave us understanding, I suppose understanding the behaviour… I think if he had had an earlier diagnosis, we would have been able to better faced the problems we had already perceived." (E)

"It absolutely totally changed our lives, we went from having been blamed from some of his behaviours to having a child who actually has a disability and it meant that you could explain what was going on to these people, and a lot of people with a decent explanation can understand at least some of it… he became eligible for support from school, although we still had to fight for that." (F)

"I think after the diagnosis, I think there definitely is a difference in myself in how I deal with other people, in the respect that when you don't have a diagnosis you do question yourself cause you get people say 'it's your parenting, go on a parenting course' and you do think well 'is it my parenting,

did I do that?' sort of thing. Em and then once you've got that diagnosis, yeah they may say it's your parenting and everything like that but I know myself that it's not." (H)

"It has changed my life enormously, I know it's just a label, but now when his behaviour was kicking off I was able to say, look he can't help it…. He has had full time support since then and it's been a huge achievement. It had a huge impact." (G)

Accepting their child

There was some discussion from the mothers on coming to terms with their child's diagnosis and accepting their child for who he is, this included changing their expectations of them to be like children who do not have Autism:

"I do feel we had to grieve for the boy we didn't have, and adapt to the boy we have now. I don't mean that we don't love him any less but it isn't what I planned." (H)

"The psychologist that I had was absolutely incredible, essentially, he said you have got to reduce your expectations of your son. For example I had an issue with him eating, he just wouldn't use crockery, he said you've got to accept that he's just not going to do it. He was almost quite firm with me really and said you know you are going to have to change your expectations and of course once did, it made life a lot easier." (F)

"I think I have had expectations that were wrong and I accept that. I always wanted a child that I could take to local things to. I had presumptions, I presumed how he would be, and that in itself was damaging to me and that made me quite upset really." (E)

"There has been a lot of time wishing he achieving what I anticipated what a child was going to be like." (G)

Support after the diagnosis

All mothers discussed the support they received or expected to receive when their child was diagnosed, with mixed responses. With all mothers in this group there was a sense of entitlement in that now that it was acknowledged that their children need help, they should be receiving help. One mother felt that she did begin receiving support and acceptance from her son's school and that she was being taken seriously: "*well in terms of school, they acknowledge now that I can't help an awful lot of what he does and they acknowledge now that his learning is very very different from other people.*" *(E)*

One mother, who had only just received the diagnosis prior to the interview, was unsure about what help to expect: "*I'm hoping that now we have a diagnosis we will learn about where the support is, right now we don't know where to even start to get information on support.*" *(H)*

However, two of the mothers felt that getting the support for her son was another struggle after the diagnosis:

"*it was really good because it was such a relief, because the stress lifted. But then you had to start fighting with education system and the health system, social services. So the stress was all built up again.*" *(G)*

"*[My son] became eligible for support at school but he wasn't getting it, he was also bullied terribly. We were very very fortunate as there was a team called autism response service and they helped us push for help from school.*" *(E)*

HEALTH

Physical Health

When discussing health two of the mothers briefly mentioned that they felt the diagnosis process had impacted on their physical health:

"I've developed over the course of, I don't know how long, fibromyalgia and obviously [the fight for a diagnosis] clearly impacted, for me it was a relief when we got it. Everywhere hurts, and this has meant I have had to take loads of time off work. And I suppose it just massively impacted on our lives." (F)

"I had had IBS since I was about 12 but I ended up seriously ill with it at this time [when going through the diagnosis process] and had treatment for chronic pain." (E)

Mental Health

All mothers in the diagnosed group discussed how their experiences of raising their child and the responses from other people had impacted on their mental health, particularly stress and depression. Three of the mothers say they have been depressed due to the stress they have experienced as a result of raising their child:

"It honestly, the last eight and a half years have been the most horrendously stressful of my entire life. I was just forever having to battle and he was like a little lunatic. So the stress has been absolutely horrendous. I have just finished a course of CBT myself because I was depressed, I just couldn't cope. My child was seen as promising. I was getting judgements from school, from family from friends, my dad, the stress has been horrendous." (E)

"I suffered quite badly from depression, and I think that was a direct consequence of being criticised by every single day. And not having the faintest idea of what to do about it." (F)

"And just got very depressed and very very down. I am now on medication for this and will be on it for the rest of my life." (G)

The other mother did not suffer from depression but she did feel her emotional wellbeing was affected and how she understood how some mothers of children with Autism get depressed: *"I haven't had to see the doctor for depression but I understand why people would in my position*

because of the stress. At times it can be incredibly stressful, because you don't get a break, no respite. I can't just sit down and chill out. I think I've coped with mine by seeking out my wonderful friends and talking about it. I have amazing support." (H)

QUALITY OF LIFE

Social Life

When the topic of general quality of life arose each mother discussed how their friendships and relationships have suffered because of the extra struggles they have in raising a child with autism, for example, they did not have the freedom to enjoy a social life, some because they found they are not able to leave their child and some because they felt other people were not tolerant and understanding of their child:

"I think my friendships have suffered, I love my friends to bits and I am sure they feel that I am a good friend but you can't always just talk over the telephone or email, you have to see them or go out or you have to take that extra step and I think that is what lets it down. I don't have the opportunity to do that." (F)

"When you go through the bad patches you lose friends and you become isolated and it is very hard to rebuild so socially our networks sort of shrank drastically. It wasn't too bad until he went to high school, then it got really bad and people couldn't deal with him so we lost friends. We stopped getting invited to places, stopped being able to invite people around, because if we did it changed things and he reacted which meant the next day at School. So yeah it was better off avoiding social situations, because we know he can't deal with them." (G)

"obviously my marriage broke down from other things as well but the stress did not help, and I think unless you know people with kids with autism as well then you do get judged an awful lot and to be honest I am the type of person who's like if you are going to judge me then I don't want to know you at all." (E)

Working Life

Three mothers discussed how raising their child has impacted on their working life, in different ways they have described how their circumstances have put extra demands on their working life and what they have done as a result of this. One mother described feeling that she was not able to give her job in social work her best as she always had to be ready to leave at a moment's notice: "*Well my son is a priority but it's meant that I get into trouble at work because I have had to take so much time off work. And when I've been on my knees health wise, I daren't take any more time off work and that can be really difficult, especially when you've had no sleep. Sometimes I have to drop everything to pick up [my son] from school because he isn't coping.*" (F)

One mother talked about how she has taken a break from working to help her child: "*I left my job, I'm a primary school teacher… and I always expected to be a primary teacher for the rest of my life but I left to settle [my son] into school.* (E)

Another mother described how she changed jobs to one that is more flexible and suitable for her as a carer for her child: "*Basically I've sacrificed everything for him. I have got a job, although it doesn't bring any company, I do enjoy it and [my son] is a lot happier. And when he is 16, I won't be so old and will go back into teaching but now I've got to put him first. I know I have made the right decision.*" (G)

A Summary of the Interviews and the differences between the groups

The interviews of both groups were compared with consideration of the Transactional Model of Stress (Lazarus and Folkman, 1984). Four topics: the diagnosis, mental health and depression, physical health and quality of life were discussed.

The Diagnosis

The key difference between these groups, that one group had a diagnosis and the other did not, had a direct effect on the how the interviews where initially shaped. The group who were seeking a diagnosis talked at length about why they believed their child has autism, i.e. aspects of their child's behaviour and developmental problems. Whereas although the group with a diagnosis discussed the difficulties they were experiencing in raising their child they put more emphasis on how these problems increased when their children were introduced to the school environment.

Both groups discussed problems they have experienced in their interactions with teaching and health professionals. The mothers seeking a diagnosis discussed interactions with many health and teaching professionals over a long period of time, a lot of which they found to be frustrating and upsetting. The group of mothers who had a diagnosis for their child described at length the difficulties they experienced with the teaching profession in particular, prior to the diagnosis, when trying to get recognition that their child was having problems and that those problems were not the fault of the parents. When relating this to the Transactional model of stress and coping it is clear that the mothers are appraising their experiences as taxing and exceeding their resources and that they did not feel they had the means to deal with their stressors, their primary appraisal. In other words they did not know how to help their child, or receive help for them and their child, so they could ultimately manage their situation better.

The group of mums seeking a diagnosis have described how they expect to feel relieved when they receive a diagnosis of HFA for their child. This is because they believe they will be no longer held accountable for their child's behaviours by strangers or health and teaching professionals, and they

expect to receive formal support of some kind. Those with a diagnosis described receiving the diagnosis as a relief and a positive step. They felt it had given them, and others understanding of their child and it helped them to accept their child for who he is. The reception of formal support following the diagnosis has been mixed with two parents describing how they have struggled to get it. It seems that those seeking a diagnosis believe that the diagnosis will give them the opportunity to reappraise their stressful situation to include an evaluation of the resources they expect will open up to them and help them to alleviate some of their stresses. Those with a diagnosis have had that opportunity, and view the meaning of the diagnosis as an important resource to gain acceptance from others.

Mental Health and Depression

Both groups discussed their stress in relation to raising their child. The group of mothers who were seeking a diagnosis discussed stress in detail by describing what was causing their high levels of stress. They believed their stress was due to worrying about not being able to help their child or manage the unusual behaviours displayed by their children, their inability to connect with their child, problems they experience when trying to get a diagnosis and not having an explanation for why their child is different. With regards to the Transactional model, in their first appraisal of stress these mothers view their situation as challenging and in their secondary appraisal they feel they are being denied the resources that will help them to deal with the challenges they are experiences, for example, the explanation that comes with a diagnosis and the help and support from the child's school that will come with a diagnosis.

Three mothers who had a diagnosis for their child briefly touched on stress by attributing depression that they had to the high levels of stress they experienced. The other mother in this group said she did not have depression but understood why mothers with children with autism do get depressed.

All four of the group of mothers seeking a diagnosis for their children were taking medication for their depression and attribute this to the stress of raising their child with autism, trying to get a

diagnosis and two of these mothers also relate it to not being able to interact and engage with their child as they had expected. So it seems, even with the resources that come with the diagnosis, the parents in the diagnosed group do appraise their situation as harming their wellbeing.

Physical Health

Only one of the mothers in the group seeking a diagnosis mentioned their physical health. She blamed the stress of seeking a diagnosis as an added pressure to raising her child she believed had autism to lead to her having chest pains. Two from the group of mothers who had a diagnosis did feel that the diagnostic process was impacting on their physical health; one was suffering with fibromyalgia and one with irritable bowel syndrome.

Quality of Life

Social and working quality of life was discussed by both groups. They felt socially isolated as a result of raising their child but described its impact in different ways. Those seeking a diagnosis discussed how their child either found social events stressful or did not fit in socially. Those with a diagnosis described the breakdown of friendships and marriages which they attributed to the impact of raising their child with autism. Both groups discussed the difficulty of balancing work and raising their child, and finding appropriate childcare for their child as having impacted on their work choices.

This can also be related to the transactional model of stress and coping. Lazarus and Folkman (1984) describe what they call adaptational outcomes, which are the quality of life aspects affected by their stress appraisal. The mothers of both groups feel their life circumstances are directly affected in that their mental well being and their ability to undertake a social and working life is hindered by what they appraise as their stressful situation.

DISCUSSION

The aim of this study was to investigate the impact on mothers of raising a child whom they believe have high functioning autism or Asperger's syndrome and are seeking a diagnosis for their child, with particular emphasis on stress and quality of life. The experimental group was compared with a group of mothers who already had a diagnosis for their child. There was one independent variable that was whether or not the mother had a diagnosis of high functioning autism or Asperger's syndrome for her child and two dependant variables, mother's stress and health related quality of life.

The study initially tested two research objectives which were comparing the participant groups on their stress and health related quality of life using a representative sample of the population in question. Further investigation via semi-structured interviews of a smaller sample of participants gave insight on participant's views on their stress and health related quality of life.

The current study was able to show that raising a child whom a mother believes to have high functioning autism or Asperger's does impact on the mother's stress levels and health-related quality of life, more so than mothers who already had a diagnosis. This finding suggests that gaining a diagnosis reduces stress and health related quality of life, although these still remained at clinical levels in the mothers with diagnosed children. Participant interview data indicated that it is the process of seeking a diagnosis in particular that causes their stress and depression, and this will be discussed further in this chapter. The highlights of the results will be examined below beginning with descriptive statistics, stress, and then health related quality of life. Other considerations that have emerged from the qualitative interviews will then be reviewed. Following that the use of the mixed methods approach and the research limitations of the research will be discussed along with areas for further research.

Descriptive Statistics

This research first looked at the descriptive statistics of both the experimental and comparison group and some differences were found. The mean ages of the participants and their children whom they were seeking or already had a diagnosis of HFA/Asperger's syndrome were both significantly higher for the comparison group. This could be explained by the fact that children who have Asperger's are more likely to be diagnosed if they are older, the average age of a child to receive this diagnosis is 11 years of age (Howlin and Asgharian, 1999). The average age of the children in the comparison group (those who had a diagnosis) were 10.8 and they had received their diagnosis at a mean age of nearly 8 years old (7.9 years), this is an improvement of Howlin's findings (Howlin and Asgharian, 1999). The mean age of the children without a diagnosis was 7.8, just slightly below the average age of diagnosis in the comparison group.

The average length of time taken to receive that diagnosis was 3.1 years, this is much longer than the NAS recommendations, whereby a child should receive a diagnosis within 30 weeks from presentation of first concern (National Initiative for Autism Screening and Assessment (NIASA), 2003). There was no significant difference between the experimental and comparison group in the mean age at which ASD symptoms were first noticed in the child and the mean age of the child when the mother first sought help.

No significant differences between groups was found in the number of other children the mothers were raising, the mother's marital status, working status and their satisfaction of formal social support and informal, familial or tangible support received, therefore these were ruled out as possible mediating factors in stress and quality of life scores.

Stress

When investigating stress, the Parenting Stress Index provided a total stress score and scores for 3 sub categories: Parental Distress, Parent-Child Dysfunctional Interaction and Difficult Child for each

participant. There was also a Defensive Responding category in which all participants scored less than 10 points which allows for the assumption that all participants answered truthfully.

The Parental Distress section of the PSI/SF scored the mother's feelings of parental incompetence, stresses associated with restrictions on lifestyle, conflicts with the child's other parent, lack of social support, and depression (Abidin, 1995). The experimental group (those who did not have a diagnosis for their child) scored significantly higher than the comparison group in this category with 98 per cent in the experimental group and 80 per cent in the comparison group scoring clinically significant high scores for this category. This is much higher than previously reported findings of just over half of mothers of children with autism suffering serious psychological distress (Bromley *et al,* 2004).

When stress was mentioned in the interviews the mothers in the undiagnosed group (those who did not have a diagnosis for their child) echoed the findings of the statistical results when they described their feelings on quite a few of the Parental Distress aspects. Restrictions on lifestyle was a major topic of discussion for them with most feeling that they have been isolated from friends and familial social activities. This was because they were afraid of how their child will react or because other people have made judgements or do not understand their child's behaviour. They felt that socialising or going to work without their child has also been a problem for them as they are unable to leave their child in the care of someone else confidently, meaning that socialising was restricted to phone calls or over the internet. The diagnosed group (those who had a diagnosis for their child) also felt their social life and friendships have suffered and that there have been demands on their working life as a result of raising their child autism. They discussed the additional issue of how they were not able to be spontaneous or do everyday things like chores or errands without precise planning.

There was mixed feelings among the diagnosed group interviewees with regards to formal social support, as even though they felt they were entitled, not all felt they were receiving adequate support, and two within this group felt that any support they did receive, they had to fight to get. These results are not surprising as previous literature has already documented that finding appropriate help and support is a challenge (Hastings and Johnson, 2001, Sivberg, 2002). The

undiagnosed group of interviewees did not talk about any support that they were receiving; they did however discuss in depth the frustrations they were feeling in their interactions with teaching and health professionals. There was very much a feeling among the undiagnosed group that they were not receiving the help or acknowledgement that they were asking for and this was why they were pushing for a diagnosis for their child.

Depression was also an important topic for both interview groups. All four of the interviewees in the undiagnosed group were taking medication for depression and they all attributed their depression to the difficult process they were going through in seeking a diagnosis for their child, two blamed their depression on not being able to engage with their child in a way they felt they should. Three of the mothers interviewed in the diagnosed groups also suffered from depression, two of whom were receiving treatment for depression, one of whom discussed her depression in retrospect.

The experimental group scored significantly higher in the Parent-Child Dysfunctional Interaction subscale with 98 per cent scoring clinically high scores, compared with 82 per cent in the comparison group. The P-CDI looked at parent's perception that the child does not measure up to expectations and that interactions with the child are not reinforcing (Abidin, 1995). The diagnosed group elaborated on this further when interviewed: when they were discussing their child's diagnosis and coming to terms with the diagnosis they felt that as part of that process they had to change their expectations of their child and some even mentioned a grieving process that they had to go through, for the child that they never had. This would not have been possible for the experimental group who do not yet have a diagnosis for their child and are still seeking answers for their child's unusual behaviours and struggles.

The Difficult Child subscale of the PSI/SF analysed the behavioural characteristics of children that make them easy or difficult to manage in the parent's view. Again a high percentage of participants in both groups scored a clinically high score with 100 per cent and 89 per cent for the experimental and comparison group respectively. However this time the difference in mean scores between the groups was not significant. In the interviews there were differences between the groups in how much emphasis participants put in their description of their child's autistic attributes. Parents in the

undiagnosed group talked at length about their children's aggression, developmental problems, sensory problems and much more. These descriptions usually arose when discussing what they were going through when seeking a diagnosis and the attributes they believed illustrated that their child did in fact sit on the autism spectrum. It was discussed in much less detail by the diagnosed group, however the diagnosed group did go into detail about the problems their children were having at school as a result of their condition, such as being bullied, being aggressive at home after a day at school and getting into trouble with the teachers.

Both groups exhibited very high overall stress. Ninety five per cent of the experimental group and 84 per cent of the comparison group scored clinically significant levels of total stress in the Parenting Stress Index. Even though this is a very high percentage in both groups, the experimental group did score significantly higher. Previous research which found that raising a child leads to high stress levels for parents (Montes and Halterman, 2007, Hutton and Caron, 2005, Hutton and Caron, 2005, Rao and Beidel, 2009, Weiss, 2002, Bromley *et al,* 2004, Hall and Graff, 2011a) has been supported by the high total stress levels found in the comparison group (those who have a diagnosis for their child) in this study.

During the interviews the general topic of stress was touched on by the diagnosed group who talked about raising their child as very stressful and how this has made many of them quite depressed. The undiagnosed group went into much more detail about what they believed was making them stressed which was a combination of their child's behaviours, the reactions from others and the stress of seeking a diagnosis.

Health-Related Quality of Life

The health related quality of life of the participants was initially measured using the Health Survey Short form then further investigated in the qualitative interviews. The Health Survey looked at mental and physical health.

No significant differences were found between groups in the Physical Component Summary score, however it is worth noting that comparison group's mean score was below the average range for the general population. This finding was supported by the qualitative interviews as two of the interviewee's in the diagnosed group did believe that the circumstances in raising their child with autism lead to them having poorer physical health. They were also mirrored by the findings in a previous study that mothers with children with HFA/Asperger's syndrome indicated poorer health-related quality of life than those in a control group (Allik, Larsson and Smedje, 2006). However the impact of raising a child with autism on the interviewee's physical health was not a significant topic of conversation in the undiagnosed group and the undiagnosed group did not present low scores for the physical component summary.

The Mental Health Summary score had significant findings in that the mean score for the experimental group was significantly poorer than the mean score for the comparison group. This meant that they would be experiencing lower vitality, social function, emotional effects on role and poorer mental health. The interviews further corroborated these findings in that both groups discussed their high levels of stress and depression as already discussed.

Quality of life was discussed in great detail by both groups in that they felt they are not able to enjoy a social and working life in the way that they wished, however they had not related this to health and most did not see it as a consequence to their health, rather they saw it as a consequence of the special needs of their child whom they are raising.

Other findings

In addition to the factors within stress and health-related quality of life the qualitative interviews gave further insight into the stressors parents were facing when raising a child who had, or whom they believed had HFA/Asperger's syndrome. Both groups talked at length about how they felt dismissed by, or frustrated and upset by the teaching and health professionals they were in contact with when seeking a diagnosis. Those who had a diagnosis for their child talked in retrospect with

particular emphasis about their problems with teaching professionals at their child's school when they were seeking a diagnosis and this was a major stressor for them. This supports findings by Rose (2011) and Keenen *et al* (2009) who found that parents experience high or very high stress due to the diagnostic process. Keenen *et al* concluded that the high stress experienced by the parents was because the statutory diagnosis takes such a long time and the care and education plans do not allow for full parental participation. Peeters (1997) wrote about the conflicting views between parents and carers when the child's behaviours vary in different situations and environments. This could also be an explanation for the conflicting views between mothers and the professionals they seek help and acknowledgement from and the stress that accompanies that conflict.

The diagnosed group talked quite passionately about receiving the diagnosis for their child. They felt it was a tremendous relief and positive step forward. This contradicts the opinion of those practitioners Filipek *et al* (1999) wrote about in their systematic review who were concerned about labelling a child and the distress this might cause the parents. The mothers in the current study felt the diagnosis enabled them to explain their child's behaviours to the schools and to strangers who they felt were judging them, it also gave their children the entitlement to receive help they needed (although they were unclear as to what help they would receive). With the diagnosis the mothers were able to reduce their expectations for their child to act like others who do not have ASD and to accept their child. These findings echo those from studies which reported the feelings of relief that came with a diagnosis because it gave parents an explanation and helped them to better understand and accept autism and the limitations it imposed on their child, it also allowed them to make adaptations for the future (Midence and O'Neill, 1999, Nissenbaum, Tollefson and Reese, 2002). The current study did not support Nissenbaum *et al*'s other findings that parents experienced negative emotions such as denial, emotionality and misperceptions of the diagnosis when they received it (Nissenbaum, Tollefson and Reese, 2002). However, this study found that even though after the diagnosis the stress mothers experience is less, they still experience high levels of stress and even depression. The interviews found that following the diagnosis there can still be a fight for acceptance

and social and educational support and this is what the mothers attribute to their high levels of stress.

During the interviews the mothers who were seeking a diagnosis for their child discussed in great detail the behaviours and struggles their children were displaying, these ranged from sleep deprivation, hand flapping to sensory problems and having tantrums among others, all traits linked to autism spectrum disorder. These behaviours and traits were tremendous stressors for the parents because they were very worried for their child and were often unsure how to manage their child's behaviours or to help their child and they felt they were being judged by others for what seemed like poor parenting. These are all common stressors experienced by parents raising a child with autism (Akshoomoff, Corsello and Schmidt, 2006, Gray, 2002a, Wiggins, Baio and Rice, 2006). These mothers were in no doubt that their child was on the autism spectrum and three of the four mothers interviewed talked about how they already treated their child as if they do have autism by, for example, using applied behavioural analysis therapy with them. This is contrary to what the interviewees expressed in retrospect in a pilot study conducted by Midence and O'Neill (1999) which were difficulties and confusions in understanding their child's behaviours prior to receiving the diagnosis. It is a possibility that the inclusion criteria for the experimental group in this study, which was that a teaching or health professional has suggested to the participant that her child has HFA/Asperger's syndrome, could have helped them to begin to learn and accept autism and what it means for their child.

The Transactional Model of Stress

Lazarus and Folkman (1984) describe stress as relationship between the person and the environment that they appraise as taxing or exceeding on their resources. Both participant groups scored very high on the Parenting Stress Index, indicating that they have clinically high levels of stress. When explored further in the interviews, both groups discuss the stressors they feel lead to their high levels of stress, for example, challenging behaviours displayed by their children and an

inability to participate in a working or social life as they expect to. However the group of mothers who are seeking a diagnosis discuss additional resources they feel are being denied, such as recognition and support from schools, and an explanation to give to others when their child is displaying challenging behaviours for which they are being judged on. Those with a diagnosis describe the relief they felt when they received a diagnosis, and it seems this diagnosis gave them the opportunity to reappraise their situation and the resources available to them. This could explain the significantly lower levels of stress indicated by the group of mothers with a diagnosis. Those who are still seeking a diagnosis will not have had that opportunity for that reappraisal.

Lazarus and Folkman (1984) also discuss the adapational outcomes in their cognitive appraisal model. This includes, functioning in work and social living, morale and life satisfaction and somatic heath. Both groups describe their inability to enjoy a work and social life and both groups discuss their depression that they attribute to the stresses of raising their child and seeking a diagnosis. This seems to correlate with Lararus and Folkman's model as the mothers have attributed their on-going stressors to their poor health and quality of life.

The mixed methods approach

The mixed methods employed in this current research began with using a quantitative approach via questionnaires on a representative sample to explore the stress levels and health-related quality of life of the participant group and a comparison group. A qualitative approach via semi-structured interviews was then employed to explain those findings.

The use of two approaches was chosen as it enabled the researcher to utilise the strengths of each. For example the quantitative approach of using questionnaires which have been previously tested for validity and reliability allowed the study of a representative sample, providing generalisability and a clear and statistical comparison between groups. However it can be argued that the questionnaires are generally inadequate in their ability to provide understanding of some forms of information such as changes of behaviours and emotions (for example, the feelings of relief in the

diagnosed group that accompanied receiving the diagnosis) and could only measure what the researcher had already perceived was relevant in the study. The interviews that followed allowed a further exploration of the findings in a less rigid way while also enabling the participants to bring to the research themes that were not yet explored by the questionnaires (for example, what the participants felt was the cause of their high stress levels).

The questionnaires chosen in the current study looked at some of the participant's circumstances in quite some detail in that they covered many aspects of parental stress such as social support, parental expectations and perceptions of child's behaviours. They also covered many aspects of health-related quality of life such as psychological distress and physical limitations. Because of the detail the questionnaires went into they were able to incorporate aspects of participant's circumstances that the interviews did not touch on, such as conflicts with the child's other parent (PSI/SF – Parental Distress) and limitations in self-care (SF-12 - Physical Health Component) and the research was able to directly ask about these topics.

A key example of a topic that the interviews brought to the research which the questionnaires could not was why the undiagnosed group felt so strongly that their children were on the Autism Spectrum even though they had not yet received a diagnosis. As this was a new research area that had not yet been explored, it was not possible to create a quantitative questionnaire that would fully capture the participant's feelings. The interviews gained lengthy descriptions of the children's traits and behaviours that the mothers attributed to autism and the strong feelings in the mothers that accompanied this. There were other topics discovered in the interviews that were captured by the questionnaires: for example, a major reason the undiagnosed group attributed to their high stress and depression levels was the feeling that other people were blaming them for their child's behaviour and that they were not being taken seriously by health or teaching professionals when seeking a diagnosis for their child, these were experiences the diagnosed group were no longer going through. The finding that some of the mothers in the diagnosed group felt that the diagnosis, or 'label', was a positive thing which enabled them to have an explanation of what they and their

children were going through and to be entitled to receive formal support specifically for autism was another example.

The data collection techniques employed in the current study did not always fulfil its intended purpose as sometimes the semi-structured interviews were not able to explore the significant findings of the quantitative data. For example, the Parent-Child Dysfunctional Interaction subcategory of the PSI found significant differences between the groups with the experimental group scoring much higher. This meant that the experimental group felt more strongly that their children did not measure up to their expectations and that their parent-child interactions were not reinforcing. This was not picked up on by the semi-structured interviews that followed, although the diagnosed group did describe in retrospect a process of grieving and acceptance for their child as a result of the diagnosis. Perhaps more structured approach to the interviews would have allowed an exploration of this more directly, enabling a more detailed and specific discussion of the quantitative findings in a qualitative framework. However, this would have inhibited the exploration of the emergent themes such as 'accepting their child' and 'support after diagnosis', which the semi-structured approach allowed.

An unexpected advantage of employing the two different methods in the current study was that in some instances the information gathered was cross-verified through data triangulation. For example, the high stress levels reported by participants in each group via questionnaires was further verified by the discussion of high stress felt by the participants who were interviewed. The lower mental health-related quality of life as reported by the experimental group through the Health Survey was echoed by the higher reported prevalence of depression by the interviewees in the undiagnosed group than the diagnosed group.

Limitations of current study

One of the main challenges in this study was defining the experimental group; those who do not yet have a diagnosis of HFA/Asperger's syndrome for their child yet feel their child does have the

condition. Without a diagnosis, whether the child meets the criteria for HFA is debatable. Researchers who have previously investigated this population group tend to study them in retrospect, post diagnosis. For example one pilot study followed four families through the diagnosis process and was able to investigate the problems parents experience in understanding their child and in obtaining a diagnosis among other topics (Midence and O'Neill, 1999), however it was not possible to do this on a representative sample with the timeframe available, especially as it may take some participants years to get a diagnosis, if they do get it. For the purposes of this study, only those who believed their child had HFA/Asperger's syndrome and have had this verified by a teaching or health professional met the inclusion criteria for the experimental group and they were recruited from Autism internet forums.

The cross-sectional approach was the chosen quantitative research method to study the target pre-diagnosis population group while comparing them with those who do have a diagnosis. However, the cross-sectional approach has limitations, for example, it was not possible to measure stress and health-related quality of life as they change over time and it was not possible to establish cause and effect. The mixed methods approach applied did however, allow for a comparison between groups at the given time and a qualitative analysis of why the differences may exist.

There has been opportunity for interviewer bias influencing the data. Some theorists argue that early literature reviews can narrow a researcher's viewpoint, directing them to focus on some aspects of the data at the expense of other potentially crucial aspects, or conversely enhance sensitivity to crucial aspects of the data (Tuckett, 2005). The interview questions in the current study were developed after the analysis of the quantitative questionnaire data and after a thorough review of previously published literature on the subject area. With this mixed methods design it was not possible to avoid the literature before data collection, and therefore it may not have been possible to avoid interviewer bias.

All attempts to reduce this bias were undertaken when possible, for example, interview questions were designed as open-ended to allow for a semi-structured style of interviewing and to encourage the participants to freely express their own perspective in their own words in as much detail as they

wished. The interviews were transcribed, reviewed and coded by the researcher so the researcher was very much familiar with all of the data before analysing it.

Participant bias was also a risk. When advertising the request for participants the researcher was transparent about the aims of the research (See Appendix A), this gave the participants the opportunity to act in a way that they believed the researcher wanted them to act. However the Parenting Stress Index's subscale Defensive Responding indicated that the participants answered truthfully and the qualitative interviews not only verified the questionnaire data but provided an in-depth explanation to the data outcomes.

Another bias that may have impacted on the results is sampling bias. Participants were self-selecting via online forums and this type of recruitment is open to selection bias or self-selection bias (Kellner, 2004). For example, only those in the research population who access online forums would be able to fill in this questionnaire or those who related to the dependant variables being tested were more likely to fill in the survey. This may also be true of the interviews because those who volunteered to be interviewed would be fully aware of what the interview would be about, having previously filled in the questionnaires and read the participant information sheet. It may be the case that those who particularly wished to express their feelings of stress and poor quality of life put themselves forward for the interviews, whereby those who did not particularly relate to the research topics did not feel they had anything to contribute to the study and therefore did not volunteer. However the fact that both participant groups did give high stress scores, and that this correlates with previous studies adds to the validation of the results in this current study, and even though both groups were stressed, those seeking a diagnosis were significantly more stressed.

The fact that the socioeconomic status of the participants was not measured opens this research up to another type of bias. Some studies have found that children are more likely to be diagnosed if their parents are better educated and have greater wealth (Thomas *et al*, 2012 and Fountain *et al*, 2011). This means there is a possibility that those in the participant group of mothers seeking a

diagnosis could be of a lower socioeconomic status and women with a lower socioeconomic status are more likely to suffer from depression and mental health problems (Groh, 2007; Belle & Doucet, 2003; Pirinicci *et al*, 2008). Both participant groups expressed high levels of stress and the group of mothers seeking a diagnosis did have poor mental health-quality of life and significantly higher stress levels. However the interviews, which discussed the topics of stress and quality of life, did indicate that participants felt that it was the stressors related to seeking a diagnosis that caused their higher levels of stress. Therefore although socioeconomic status was not indicated as a possible factor by the interviews, it was also not illuminated as a possible factor by the questionnaires. Further research should include reviewing socioeconomic status in the demographics of participants as a possible mediating factor when looking at the experiences of parents seeking a diagnosis of autism for their child. .

Recommendations for Practice

This research has indicated a link between seeking a diagnosis and high stress, depression and poor mental health-related quality of life, as a result some recommendations for practice have emerged. This first recommendation is for a psychologist in the autism diagnosis team to consider the health of the parent by assessing if they are experiencing stress, depression or poor health-related quality of life. If there are such concerns then an appropriate referral should be offered to the mental health services available to them in that area, for example, Improving Access to Psychological Services that offer free psychological therapy to people with anxiety and depression throughout England.

The second recommendation is to provide information and/or referral links for the parents so they know where to go or what steps to take next in order to seek further help and support in raising their child. Support that would benefit these parents would include training in how to manage their child's behavior, social networks for parents in similar situations and respite or specialised child minding services to allow the parents time away from caring for their child. The information can be in the form of a referral pack to be given to parents when the diagnostic assessment has been communicated to them.

Areas for further research

The focus of the current study was to learn about the experiences of the mothers raising a child whom they believe has autism and compare them with the mothers raising a child with a diagnosis of autism. This is because of the assumption that the mothers were the main carers of their children (Gray, 2003). Two other population groups neglected in research are the fathers and siblings of children with undiagnosed autism who may bring very different viewpoints to research literature. Studies have found that fathers of diagnosed children tend to experience less parental stress than mothers do and that their stress is predicted by their partners stress (Gray, 2003, Hastings *et al,* 2005). Siblings of children with autism are also more likely to have emotional problems than a control group (Petalas *et al,* 2009). It would be interesting to learn how raising a child who has autism which is not yet diagnosed could influence these findings. Further research within this field would also benefit from a mixed-methods approach similar to that of this current research but with a longitudinal design, this would allow for analysis on causality.

Conclusion

Mothers who are raising children with high functioning autism/Asperger's syndrome do experience clinically high levels of stress and low health-related quality of life as reported in previously documented research and in this study. The fact that the group of participants in the current study who do not yet have a diagnosis report significantly higher stress and lower health-related quality of life than those who do have a diagnosis for their child is noteworthy. Whether or not these children do sit on the autism spectrum does not invalidate the on-going stresses these participants are experiencing, and the possible consequences of those stresses, i.e. a reduced resistance to stressors and depression. Some researchers estimate that there are approximately 157 children per 10,000 with Autism, including undiagnosed, in the United Kingdom (Baron-Cohen *et al,* 2009), which suggests that there is a large population of mothers seeking diagnoses of autism for their children and who may need the understanding and support that does come with a diagnosis. Yet this

population group is largely neglected in the literature. The highlights of this current study reveal areas where in-depth research and support systems would benefit parents, teaching and health practitioners and the children who are going through the autism diagnostic process living in the United Kingdom today.

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APPENDICES

Appendix A: participant information sheet, informed consent, a debrief statement and an invitation to participate in the qualitative interviews

Appendix B: Demographic questionnaire

Appendix C: Interview schedule

Appendix D: The 6 Phases of Thematic Analysis

Appendix E: Ethical Approval

Appendix F: Themes and subthemes

Appendix G: Interview questions

Appendix A

Mother's experiences raising a child with Autism

Information, Consent form and personal circumstances

You are being invited to take part in a research study. Before you decide to take part please take time to read the following information carefully and discuss it with others if you wish. Please ask the researcher if you have any questions or if anything is not clear and you would like more information.

 1.   **Study Title**

Mothers' experiences of raising a child with high functioning autism or Asperger's syndrome: an evaluation of stress and quality of life.

2.   **What is the purpose of the study?**

Recent studies have found that it will take an average of 2 years to get a diagnosis of autism and health professionals are often reluctant to give a diagnosis of autism or Asperger's syndrome. On average, those children with high functioning autism or Asperger's syndrome are not diagnosed until they are 11 years old. This can have a tremendous effect on the child and family. The aim of this study is to compare levels of stress and quality of life in mothers raising a child with a diagnosis of high functioning autism or Asperger's syndrome with those mothers of children who do not have a diagnosis of Autism. No studies have yet compared the stress and quality of life in mothers raising a child with autism in those with a diagnosis and those without.

3.   **What will happen to me if I take part?**

If you do decide to take part in this researcher you will simply fill in the consent form on the next page to say that you will be participating in this research then fill in the 4 questionnaires in the following pages. The questionnaires should take no more than 20 minutes of your time. The four questionnaires will focus on your personal circumstances, stress and quality of life.

The researcher would also like to interview a number of participants about their experiences about getting a diagnosis and raising a child with Autism or Asperger's syndrome. The purpose of this interview is to explore the challenges, difficulties and rewards parents are facing and their resources and needs. The interviews will be conducted via telephone and will be recorded by the researcher so the researcher can review the conversation at a later date. No participants who have answered the questionnaires are obliged to be interviewed. However if you would like to be interviewed please leave your email address in the space provided in the Debrief or email Ashlee Mulimba at ashlee2.mulimba@live.uwe.ac.uk.

When all the data has been collected by the researcher the researcher will compare the information of those mothers who have a diagnosis for their child and those who do not.

4.   What are the possible disadvantages and risks to taking part?

There are no disadvantages to taking part; all information given to us will be completely confidential.

5.   Will my taking part in this study be kept confidential?

All information that is collected about you during the course of this research will be kept strictly confidential and will not contain any of your identifiable details. Only the researcher and two research supervisors from the University of West of England Health Psychology faculty will have access to the data you give us.

6.   What will happen to the results of the study?

The completed research will be written up with a view to be published. If you would like to be informed of the results of the study and any publications then please contact Ashlee Mulimba at ashlee2.mulimba@live.uwe.ac.uk and leave your email address. Your contact details will be kept apart from any other information you provide to us should you decide to partake in this research.

It is expected that this research will be completed in September 2012.

7.   Who is organising and funding the research?

The research is organised by Ashlee Mulimba, a trainee Health Psychologist and Doctoral Student at the University of the West of England. All expenses are covered by Ashlee Mulimba.

8.   Who has reviewed this study?

This study has been reviewed and approved by the University of West of England Postgraduate Research Committee and Faculty Ethics Committee.

9.   Contacts for further information

If you would like more information please contact Ashlee Mulimba on ashlee2.mulimba@uwe.ac.uk or Dr James Byron-Daniel on james.byron-daniel@uwe.ac.uk.

Consent

- ☐   I confirm that I have read and understood the information above and that I agree to take part in the study
- ☐   I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my medical or legal rights being affected.

Debrief

Q34

☐

Thank you for taking the time to complete this survey, your help is very much appreciated.

If you would like to take part in the interview regarding your experiences in raising your child and obtaining a diagnosis please leave your email address and/or other contact details below. Alternatively you can email Ashlee Mulimba on ashlee2.mulimba@uwe.ac.uk.

Q35

☐

The aim of this study is to compare levels of stress and quality of life in mothers raising a child with a diagnosis of high functioning autism or Asperger's syndrome with those mothers of children who do not have a diagnosis of Autism. No studies have yet compared the stress and quality of life in mothers raising a child with autism in those with a diagnosis and those without.

If you would like to be informed of the results of the study and any publications then please contact Ashlee Mulimba at ashlee2.mulimba@uwe.ac.uk and leave your email address. Your contact details will be kept apart from any other information you provide to us should you decide to partake in this research.

If at any point you wish to withdraw from this study please contact Ashlee Mulimba on ashlee2.mulimba@uwe.ac.uk with the date and approximate time you filled in the survey and your information will be removed and deleted immediately. You will not be required to give a reason and your medical care or legal rights will not be affected.

 If you have any further questions please do not hesitate to contact Ashlee Mulimba on ashlee2.mulimba@uwe.ac.uk or James Byron-Daniel on james.byron-daniel@uwe.ac.uk.

 Many thanks,

Ashlee Mulimba

Demographic Questionnaire

Has your child been diagnosed with high functioning autism or Asperger's syndrome?

- ○ Yes
- ○ No

If Yes, how old was your child when he/she was diagnosed?

○ Aged 3 or younger	○ 11
○ 4	○ 12
○ 5	○ 13
○ 6	○ 14
○ 7	○ 15
○ 8	○ 16
○ 9	○ 17
○ 10	○ 18 years or older

If No, has a teaching, health or social services professional suggested to you that your children may have high functioning autism or Asperger's syndrome?

- ○ Yes
- ○ No

What is your Age?

☐

What is your child's Age?

How many other children have you got and what are their ages?

Are you working (either paid or voluntary) at the moment?

- ○ Yes
- ○ No

☐ **Do you have a husband or partner who lives with you at home?**

- ○ Yes
- ○ No

☐

How old was your child when you first thought something was wrong?

○ Aged 2 or younger	○ 9
○ 3	○ 10
○ 4	○ 11
○ 5	○ 12
○ 6	○ 13
○ 7	○ 14
○ 8	○ 15 or older

☐

How old was your child when you first sought help?

○ Aged 2 or younger	○ 9
○ 3	○ 10
○ 4	○ 11
○ 5	○ 12
○ 6	○ 13
○ 7	○ 14
○ 8	○ 15 or older

☐ **Does your child have any of the following associated conditions/symptoms:**

- ☐ Attention Deficit Hyperactivity Disorder
- ☐ Aggressive behaviour
- ☐ Self-harm behaviour
- ☐ Epilepsy
- ☐ Bed wetting
- ☐ Soiling

Have you had any contact with the following services through your child?

- ☐ Community Paediatrics
- ☐ Child and Adolescent Mental Health Services
- ☐ Social Services
- ☐ Health Visitor
- ☐ School Nurse/Doctor

☐

Please list the people/services whom you count on when you are in need of emotional/tangible support, by giving their relationship to you.

Q15
☐

In a scale of 1-10, where 1 is very dissatisfied and 10 is very satisfied please rate your overall satisfaction with the following

	1.Very dissatisfied	2	3	4	5	6	7	8	9	10. Very Satisfied
satisfaction from services	○	○	○	○	○	○	○	○	○	○
satisfaction of emotional/tangible support received	○	○	○	○	○	○	○	○	○	○

Interview Schedule

Interview	Participant	Date and Time	Diagnosis Status	Interview Status
1	R_byLvNOXTiDYO3qY	3rd September – 10:30am	Non- Diagnosed	Transcribed
2	R_71yyMezF9J1TDs8	3rd September – 12:30pm	Diagnosed	Transcribed
3	R_4NlTR6qLXNkJlU8	12th September – 8pm	Non- Diagnosed	Transcribed
4	R_2nUUMR3T4MG9jMw	13th September – 10am	Non- Diagnosed	Transcribed
5	R_6XQMnxMPKMx7NGs	13th September – 6:30pm	Diagnosed	Transcribed
6	R_50Xb5wBOhOQxhPe	19th September - 1.30pm	Diagnosed	Transcribed
7	R_djv9VpwP6cEJFYw	19th September - 3pm	Non- Diagnosed	Transcribed
8	R_3UG6Q1b9qJVKrNq	19th September - 8pm	Diagnosed	Transcribed

Phases of Thematic Analysis

Phase	Description of the process
1. Familiarising yourself with your data:	Transcribing data (if necessary), reading and re-reading the data, noting down initial ideas.
2. Generating initial codes:	Coding interesting features of the data in a systematic fashion across the entire data set, collating data relevant to each code.
3. Searching for themes:	Collating codes into potential themes, gathering all data relevant to each potential theme.
4. Reviewing themes:	Checking in the themes work in relation to the coded extracts (Level 1) and the entire data set (Level 2), generating a thematic 'map' of the analysis.
5. Defining and naming themes:	Ongoing analysis to refine the specifics of each theme, and the overall story the analysis tells; generating clear definitions and names for each theme.
6. Producing the report:	The final opportunity for analysis. Selection of vivid, compelling extract examples, final analysis of selected extracts, relating back of the analysis to the research question and literature, producing a scholarly report of the analysis.

Source: Braun, V. & Clarke, V. (2006) 'Using thematic analysis in psychology' Qualitative Research in Psychology, 3, 77101.

University of the West of England, Bristol
Faculty of Health & Life Sciences
Research Governance
Project Certificate

Project Details   Overall approval status for | HLS10-2265 | is | ***APPROVED*** |

Project Title:
STRESS AND HEALTH-RELATE
QUALITY OF LIFE IN MOTHERS
SEEKING A DIAGNOSIS OF HIG
-FUNCTIONING AUTISM FOR TH

Project Area/Level: /

Proposed Start/End Dates: 05-12-2011   /   01-06-2012

Chief Investigator: Ashlee   Mulimba

Supervisor/Manager: James   Byron-Daniel

☑ <-Approval Lock should be checked

Section Status: Approved

Ethics   Ethics Not Required? ☐   or Previous Approval? ☐

Supervisor/Manager Status/Approval: ☑

Ethics Scrutineer Status/Approval: ☑

Ethics Chair Status/Approval: ☑

UWE Ethics Comm Status/Approval: ☐

Ethics Section Status: Approved

Health & Safety   Low Risk? ☑   or Previous Approval? ☐

Supervisor/Manager Status/Approval: ☑

H+S Scrutineer Status/Approval: ☐

H+S Chair Status/Approval: ☐

H+S Section Status: Approved

Genetic Modification   No use of GM Organisms?: ☑

Supervisor/Manager Status/Approval: ☑

GM RA Lead Worker Status/Approval: ☐

GM Chair Status/Approval: ☐

GM Section Status: Approved

Animal Care & Husbandry   No Involvement of Animals?: ☑

Supervisor/Manager Status/Approval: ☑

Animal Care Chair Status/Approval: ☐

THEMATIC ANALYSIS – Themes and subthemes

Parents who are seeking a diagnosis for their child

The Diagnosis

Quality of Life

Belief child has autism

Interactions with professionals

Expectations that things will change

Stress

Mental Health

Depression

Parents who have a diagnosis for their child

Struggles before diagnosis

The Diagnosis

Social support after diagnosis

Receiving diagnosis

Accepting child

Noticing the child has a problem

Problems with school

Trying to get recognition of condition

Mental health

Quality of Life

Physical health

Health

Social Life

Working Life

Appendix F

<div align="right"><u>Appendix G</u></div>

Interview questions

- Can you tell me about your experiences in getting/searching for a diagnosis for your child?
- How has having a diagnosis changed your life/how do you believe getting a diagnosis will change your life?
- If stress has impacted on your life and raising of your child/children, how?
- What helps you cope?
- How has raising your child impacted on your health?
- How has raising a child impacted on your quality of life?

Chapter 2

Systematic Review: Are Motivational Interviewing (MI) based interventions effective at improving health behaviours for people with Diabetes mellitus?

ABSTRACT

Purpose

Motivational interviewing [MI] is a patient-centred counselling style that has been developed for the management of a wide variety of health care treatments including long term conditions such as Diabetes. Diabetes is a condition that requires life-long management and self-care in order to promote quality of life and reduce the risks of additional serious health problems. The aim of this systematic review is to assess whether motivational interviewing based interventions are effective at improving health behaviours in people with Diabetes mellitus type 1 and 2 by encouraging practices such as smoking cessation, healthy eating, exercise uptake, alcohol reduction and weight, blood glucose, blood pressure and cholesterol management.

Methods

Electronic databases were searched for articles which researched the use of MI and diabetes self-management between 1966 and March 2010. In total 112 titles were identified, of those 24 abstracts were identified as potentially relevant to this review, i.e. studies which researched the effects of MI on diabetes self-management on an adult population. No studies were excluded on the basis of their methodology. Using a data extraction sheet, eight studies were identified as relevant once full articles were examined.

Results

Eight articles examined the use of MI to improve self-management behaviour in people with diabetes. Positive results in health behaviour improvement were reported which included reduced smoking, improved blood glucose, improved diet and weight management. These were found in four of the studies. Studies varied in quality, four were methodologically weak due to small sample sizes,

lack of clarity of scoring measurement tools, limited use of valid measurements and reported inclusion/exclusion criteria.

Conclusions

Although findings suggest a positive link between MI and diabetes self-management, quality research in this area is limited. More robust research such as randomised control trials with larger sample sizes are needed in this area before recommendations for its use can be made.

INTRODUCTION

Diabetes mellitus is a condition which occurs when the amount of glucose in the blood is too high because the body cannot use it properly (Diabetes UK, 2010). Since 1996 the number of people diagnosed with diabetes has increased from 1.4 million to 2.6 million (Diabetes UK, 2010), effective diabetes management can reduce the incidence and progression of serious health problems related to diabetes such as blindness (Ockrim and Yorston, 2010), heart disease (Patel et al, 2008), kidney failure (Moorish et al, 2001), stroke (Jeerakathil et al, 2007) and depression (Katon et al, 2004). One in 10 people admitted to hospital has diabetes and it has been estimated that in the UK, 10 per cent of the NHS budget is spent on diabetes care (Department of Health, 2006). Effective diabetes treatment can reduce the incidence and progression of diabetes lead to better quality of life for the patient and lower costs for health care services (NICE, 2003).

Treatment of diabetes involves lowering blood glucose and the levels of other known risk factors that damage blood vessels; for example, tobacco cessation is important to avoid complications such as cardiovascular disease (WHO, 2009). Blood glucose control is considered effectively managed if levels are at 7 per cent or below and can be managed through insulin for Type 1 diabetes and oral medication for Type 2 diabetes (National Institute for Clinical Excellence, 2009). Other management methods would include blood pressure control, foot care and blood lipid control to regulate cholesterol levels and alcohol reduction. Maintaining a healthy lifestyle is also key as a health improvement tool for diabetes, this includes achieving and/or maintaining a healthy body weight, being physically active, maintaining a healthy diet which includes 3-5 servings of fruit and vegetables a day and reduced saturated fat and sugar intake (WHO, 2009).

Medical model styled techniques such as direct questioning, persuasion and giving advice have proven to be limited in their effectiveness when managing chronic illness such as diabetes in a patient (Anderson and Funnell, 2000). Motivational Interviewing [MI], an approach that was initiated by Miller (1983) and developed by Miller and Rollnick (1991), focuses on providing opportunities to clients so that they can assess for themselves what is important to change, possible to change and how that change can be achieved (Welch et al, 2006). Its approach is described as "a collaborative, person-centered form of guiding to elicit and strengthen motivation for change" by Rollnick and Miller (2009). Motivational Enhancement Therapy [MET] is a time-limited four-session version of this where the therapist uses a more systematic approach and employs "motivational strategies" to mobilise the client's own change resources (Miller et al, 1992).

According to Rollnick et al (2008) the specific and trainable behaviours characteristic of a motivational interviewing style are: seeking to understand the person's frame of reference, particularly through reflective listening, expressing acceptance and affirmation, eliciting and selectively reinforcing the client's own self motivational statements, expressions of problem recognition, concern, desire and intention to change, and ability to change, monitoring the client's degree of readiness to change, and ensuring that resistance is not generated by jumping ahead of the client and affirming the client's freedom of choice and self-direction. These basic styles can be learned and applied in clinical settings by a range of health care professionals and are described as a useful tool in reducing clinician frustration with 'noncompliant' patients (Welsh et al, 2006).

Several systematic reviews have assessed the effectiveness of MI for different health behaviours with mostly positive results. For example, Dunn et al (2001) demonstrated that MI is an effective substance abuse intervention method when used by clinicians who are non-specialists in

substance abuse treatment. Rubak et al (2005) looked at MI in different areas of disease and found that MI was effective in combined effect estimates for body mass index, total blood cholesterol, systolic blood pressure, blood alcohol concentration and standard ethanol content, but not significantly effective in cigarettes per day and for HbA1c levels. Knight et al (2006) looked at MI in physical health care settings and the majority of studies they found presented positive results of MI on psychological, physiological and life-style change outcomes although reported lack of strong evidence.

One systematic review and meta-analysis looked at randomised controlled trials of psychological interventions to improve glycaemic control in patients with type 2 diabetes and found improvements in long-term glycaemic control and psychological distress but not in weight control or blood glucose concentration in people who receive psychological therapies (Ismail et al, 2004). Another review looked at MI to improve blood-glucose control in childhood diabetes (Gregory & Channon, 2009) and found that MI is useful when working with teenagers. They found that this approach seemed effective in facilitating healthier approaches to diet and exercise and glycaemic control in young people with diabetes. No review articles have been found that have systematically examined the effectiveness to which MI can improve health behaviours in adults with diabetes, such as weight management, physical activity, healthy eating, smoking cessation, foot care, medicine adherence, alcohol reduction and glucose and blood pressure monitoring and control and blood lipid control.

This review aims to systematically review studies using MI as an intervention to improve health behaviours in people with Diabetes Mellitus as outlined by the World Health Organisation (2009) and National Institute for Clinical Excellence (2009). This will involve reviewing:

- Lifestyle and non-pharmacological self-management techniques including healthy eating, glucose monitoring and control, blood pressure monitoring and control, blood lipid control,

weight loss, physical activity, foot care, smoking cessation, alcohol reduction and medicine

adherence

- The quality of the research into this topic area

METHODOLOGY

Selection of studies for inclusion

Keyword search was conducted on the following databases: Cochrane Central Register of Controlled Trials, Medline 1966–March 2010, Embase 1980-March 2010, PsycInfo 1974 – March 2010, Science Direct, Google and www.motivationalinterviewing.org. CINAHL, Dissertation Abstracts International and NHS Evidence in Health and Social Care were searched for studies that had not yet been published, their authors were contacted for more information on their research. References of similar reviews and potential research studies were hand searched for relevant articles.

Keywords included 'motivational interviewing' and 'diabetes'. The search was then expanded to include papers that used MI but did not contain MI in the title or abstract, keywords such as 'rolling with resistance' and 'Motivational Enhancement Therapy' were used. Other search terms for Diabetes were used such as 'IDDM', 'NIDDM', 'glycaemic control' and 'A1c'. MeSH terms were also identified and searched. Only articles printed in the English language were used.

Search selection

The initial search produced 464 articles. Once duplicates were removed there were 112 remaining articles. The first stage of the selection process involved reviewing the abstracts for the following inclusion criteria:

- Population: Adults (aged 16 and over) with Type 1 or 2 Diabetes

- Intervention: Motivational Interviewing

- Comparison: No intervention or a non-MI based intervention

- Outcomes: Improvement in health behaviours including one or more of the following: weight management, physical activity, healthy eating, smoking cessation, medicine

adherence, alcohol reduction and glucose, blood pressure and cholesterol monitoring and control.

- Types of study: Both randomised controlled trials and non-randomised controlled trials were included in this study.

The search selection is demonstrated in Figure 1.

[Insert Figure 1]

Paper retrieval

Twenty four abstracts were identified as potentially relevant to this review from the 112 abstracts identified and full articles were obtained. To ensure selection bias was at a minimum the articles were assessed by 2 researchers separately with the understanding that any disagreements would be settled by another impartial researcher, at both stages of the selection process.

Quality assessment

The quality of the studies was assessed using Effective Public Health Practice Project Quality Assessment Tool (2003) as it assesses internal and external validity by rating the following criteria for each study: selection bias, allocation bias, confounding, blinding, data collection methods, withdrawals and dropouts, statistical analysis and intervention integrity. Using this quality assessment tool the studies are rated weak, medium or strong. Content validity and test retest reliability of this assessment tool had already been established (Thomas et al, 2001; Thomas et al, 2004).

Data synthesis

A summary of the data formulated to provide a brief account from the studies was conducted using a detailed data extraction sheet, which was then tabulated identifying key characteristics of the study (Table 1 and 2 in appendices). Due to the differences of the outcome

measures, study location and participant profiles between the studies it was not considered statistically appropriate to combine the outcomes in a meta-analysis (Eysenck, 1994).

RESULTS

Summary of Studies

Tables 1 and 2 show a summary of the main characteristics of each study included in the review. Eight published articles were found to test MI on the health behaviours of people with diabetes, 4 of which were considered methodologically strong (Ismail et al., 2008; Smith West et al, 2007; Hokanson et al, 2006 and Brug et al, 2007) and 4 methodologically weak (Smith et al, 1997; Calhoun et al, 2010; Rubak et al, 2009 and Britt et al, 2008).

Of the 8 studies selected for this review, 6 were randomised control trials, 4 of which were considered methodologically strong (Ismail et al., 2008; Smith West et al, 2007; Hokanson et al, 2006 and Brug et al, 2007), one was a pilot and therefore underpowered (Smith et al, 1997) and one compared follow-up data only (Rubak et al, 2009). Of the remaining 2 studies, one was a cohort (one group pre-post) study (Calhoun et al, 2010) and one was an experimental design (Britt et al, 2008). Three of these studies tested exercise/physical activity (Ismail et al., 2008; Smith et al, 1997 and Calhoun et al, 2010), 5 tested dietary intake (Ismail et al., 2008; Smith West et al, 2007; Brug et al, 2007; Smith et al, 1997 and Calhoun et al, 2010), 2 tested smoking behaviour (Brug et al, 2007 and Rubak et al, 2009), 2 tested alcohol intake (Ismail et al., 2008 and Rubak et al, 2009), 7 tested glucose control (HbA1C) (Ismail et al., 2008; Smith West et al, 2007; Hokanson et al, 2006; Brug et al, 2007; Smith et al, 1997; Calhoun et al, 2010 and Britt et al, 2008), 3 tested cholesterol (LdL) (Ismail et al., 2008; Hokanson et al, 2006 and Britt et al, 2008) and 3 tested blood pressure (Ismail et al., 2008 and Hokanson et al, 2006). These have been summarised in Table 3.

Three of the randomised control trials outlined their methods of randomising (Ismail et al., 2008; Hokanson et al, 2006 and Rubak et al, 2009) and two reported a calculation of sample size

requirement based on power and were able to achieve their targeted sample sizes (Ismail et al., 2008 and Brug et al, 2007).

[Insert Table 1, 2 and 3]

Measures

Exercise/physical activity and dietary intake was measured through a variety of methods including established questionnaires (Ismail et al., 2008; Smith West et al, 2007 and Brug et al, 2007), food diaries (Smith West et al, 2007), and self-report calorie intake and physical activity of which the scales are unclear (Smith et al, 1997 and Calhoun et al, 2010). Brug et al (2007) assessed and confirmed validity of their questionnaire of choice: the Food Frequency Questionnaire. Ismail et al (2008) reported reliability and validity of their chosen measure: the Summary of Diabetes Self-Care Activities scale as assessed by Toobert et al (2000). This validated scale was also used to measure Alcohol in 2 studies (Hokanson et al, 2006 and Rubak et al, 2009) and smoking in one study (Rubak et al, 2009). Smoking was measured in another study through participant self-reporting. Body Mass Index and waist circumference measures were used to assess weight management (Ismail et al., 2008; Smith West et al, 2007; Hokanson et al, 2006; Brug et al, 2007 and Smith et al, 1997) and measurement guidelines were clearly detailed in one study (Smith West et al, 2007).

Inclusion/Exclusion criteria

Four of the selected studies outlined their participant inclusion/exclusion criteria (Ismail et al., 2008; Smith West et al, 2007; Hokanson et al, 2006 and Smith et al, 1997). Inclusion criteria included factors such as ability to participate in exercise programmes (Smith West et al, 2007 and Brug et al, 2007), were overweight or obese (Smith West et al, 2007). Exclusion criteria included factors such as pregnancy (Ismail et al., 2008 and Smith West et al, 2007) and psychiatric illness (Hokanson et al, 2006). Only one study included participants with Type 1 diabetes only (Smith West et al, 2007), 6 included participants with Type 2 diabetes only (Smith West et al, 2007; Hokanson et

al, 2006; Brug et al, 2007; Smith et al, 1997; Calhoun et al, 2010 and Rubak et al, 2009) and one study included both Type 1 and 2 diabetes (Britt et al, 2008).

FINDINGS

Blood glucose control

HbA1C measurements were taken to assess blood glucose control in 7 of the studies (Ismail et al., 2008; Smith West et al, 2007; Hokanson et al, 2006; Brug et al, 2007; Smith et al, 1997; Calhoun et al, 2010 and Britt et al, 2008), 2 of which reported findings that MI was significantly effective in reducing patient's glucose levels (Smith West et al, 2007; Smith et al, 1997). These 2 studies were similar in that they were both set in USA, were primarily studying the effects of MI on weight loss in women with Type 2 diabetes and both were conducted by the same first author. Smith et al (1997) demonstrated a significant difference at their four month follow up, Smith West et al (2007) was able to demonstrate significant difference between groups at six months, however this effect was not retained at twelve and eighteen months follow up. Studies by Ismail et al (2008) and Hokanson et al (2006) found better improvement in blood glucose with the MI intervention groups, and Ismail et al (2008) found an even better improvement with a combination of Cognitive Behavioural Therapy and MI, however the differences between groups were not significant. One study reported some adverse reaction in the intervention group, with 8 of the participant group (n=26) exhibiting worse blood glucose control (Calhoun et al, 2010). Hokanson et al (2006) reported no significant differences between groups in blood glucose control, however their intervention focused on smoking reduction with MI and not directly on glucose management.

Dietary Intake

Motivational Interviewing was found to have a significant effect on dietary intake in 2 studies (Smith West et al, 2007 and Brug et al, 2007). Smith West et al (2007) assessed self-monitoring food diaries submitted by their participants which assessed calories and fat gram intake, they scored their MI group significantly better at their 6 (0.003), 12 (0.003) and 18 month (0.005) follow ups, although they did not outline their scoring technique for this measure. Brug et al (2007) used the food frequency questionnaire on intake of saturated fat, fruits and vegetables. They found significant improvement in their MI group compared to their control group in saturated fat at 5-6 months post baseline, no significant differences were found in fruit and vegetable intake between groups.

Weight Loss

Only one study was successful in evidencing MI as a positive tool in influencing weight loss in women with diabetes (Smith West et al, 2007). Smith West et al (2007) demonstrated significant weight loss for both their intervention group and their control group, with significantly more weight loss in their MI group at 6 months (≤ 0.01), 12 months (≤ 0.02) 18 months (≤ 0.04). Smith West et al (2007) also compared USA white population and African American population's weight patterns at each follow up for both treatment conditions and found that white women weighed significantly less than baseline at each follow up (all $p < 0.0001$).

Smoking

Two studies investigated MI as an intervention for smoking reduction in adults with diabetes (Hokanson et al, 2006 and Rubak et al, 2009). Hokanson et al (2006) who focused their study on smoking reduction measured participant self-report on a 7 day point prevalence of smoking at 3 and 6 months follow up by asking participants if they smoked a cigarette in the last 7 days and found no significant differences between groups. At 3 months they recorded a significantly lower percentage

of participants who self-reportedly smoked daily in the intervention group (p=0.048). Rubak et al (2009) who used the Summary of Diabetes Self-Care Activities scale noted no significant differences in numbers of participants who smoked between groups in their 1 year follow up. These findings were not compared to baseline data and any smoking cessation interventions used were not discussed.

Cholesterol, blood pressure, alcohol and exercise/physical activity

No studies were able to demonstrate MI as having an affect on cholesterol, blood pressure, levels of alcohol consumed or levels exercise/physical activity undertaken in people with diabetes.

DISCUSSION

This research identified and reviewed 8 articles examining the use of MI to improve health behaviours in people with diabetes. These studies variably looked at blood glucose control, cholesterol, blood pressure, exercise/physical activity, diet, weight management, smoking and alcohol. Six of these studies were randomised control trials, four of which were considered methodologically strong by the researchers using an external and internal validity tool, one was underpowered and one compared follow up data between groups. Of the 2 remaining studies, one was a cohort study and one an experimental design both were underpowered.

Motivational interviewing is a client-centred method health practitioners can draw on to enhance motivation to change in their patients and fits very well with the move towards client-centred care in diabetes (National Institute for Health and Clinical Excellence, 2009). It is encouraged as a method health care professionals are skilled at when promoting health behaviours by the Department of Health (Department of Health, 2009). However, this systematic review has found little evidence to support its current popularity.

Half of the studies reviewed found positive and significant effects of MI for adults with diabetes in only 4 of the 8 health behaviour topics investigated. These behaviours were smoking, blood glucose, diet and weight management. This further supports the evidence yielded by previous systematic reviews that MI is an effective treatment method (Dunn et al, 2001; Rubek et al, 2005; Knight et al, 2006 and Gregory & Channon, 2009). However this evidence was inconsistent, for example, only two studies, both randomised control trials, found a link between MI and positive blood glucose management, although one of these studies was underpowered and both of these studies were written by the same first author. Only one study found significant findings for weight loss and one for smoking. Two studies found significantly better dietary intake in participants in the MI groups than the control groups, however one of these studies did not report their scoring

technique. Overall, where a positive link between MI and improvement in health behaviours in people with diabetes was found, more studies were found to suggest no statistically significant effect. Little research that investigated the effects of MI on health behaviours of people with diabetes was found hence it is inappropriate to draw conclusions on any affects found with an MI intervention.

Summary

An extensive literature search for evidence on MI and health behaviours in people with diabetes was conducted, yet only 8 relevant studies were found. These studies varied in outcomes tested, populations tested, measurements used, concurrent treatments and clarity of methodology and decisive conclusions cannot be drawn from half of these studies due to methodological concerns such as small sample sizes, lack of clarity of scoring measurement tools, limited use of valid measurements and reported inclusion/exclusion criteria.

Most studies provided an adequate account of the MI and quality control measured used, however considering the variety of population groups studied and outcomes measured, no articles mentioned modifications made, if any, to suit specific needs of their unique programme. Only half of the studies adequately discussed the concurrent or control treatments provided to the patients in their programmes, and not all outlined the MI training provided to the interventionists, thus omitting information on possible confounding factors.

Strengths and limitations of this study

The limited number of studies that were found to investigate MI and diabetes health behaviours could be due to factors such as publication bias, whereby smaller studies or studies that have not produced positive results will not have been published and are therefore inaccessible to the researcher (Song et al, 2010). Several dissertation abstracts were found by the researchers and

attempts were made to contact the author for more information but were not successful. Other potential research articles could have been missed by excluding non-English papers. To illuminate any further bias two researchers were involved in both stages of the paper selection process and using a data extraction sheet both researchers identified and discussed the strengths and limitations of each research article. The researchers included all studies that investigated the relevant variables, regardless of the strength of the study, this was to ensure that any noteworthy findings were not missed. There were too few studies which varied in patient groups and outcomes tested therefore it was not considered appropriate to conduct a meta-analysis (Eysenck, 1994), when further work in this area is published this may become appropriate in an updated version of this review.

Implications for further research

When investigating available research into MI and its effects in physical health care settings Knight et al (2006) found positive results, but were not able to draw strong conclusions due to the weakness of the studies involved. This systematic review is similar in that it is unable to draw strong conclusions from its findings; of the 4 stronger studies found by this review, only 2 were able to demonstrate that motivational interviewing has positive effect health behaviours in people with diabetes.

More high quality trials and research is needed in the area of MI and health behaviours of people with diabetes, though this is a difficult variable to test. Although quality control measures were in place in 4 of the studies it can still be very difficult to standardise the 'styles' adopted and tested by the interventions and therefore difficult to measure what aspects of motivational interviewing are effective. It may be more appropriate to test a standardised MI training programme and the outcomes for the clients or patients of the health care professionals who are trained. This was the method adopted by two of the studies found (Brug et al, 2007 and Rubak et al, 2009). More research is needed that will also utilise standardised and validated measurements, larger sample sizes and

power calculations and are clearer in their use of MI in order to fully appreciate the benefits of Motivational Interviewing.

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APPENDICES

Appendix A: Flow chart showing the retrieval process of studies included in the systematic review

Appendix B: Main characteristics of the four stronger studies included in the systematic review

Appendix C: Main characteristics of the weaker studies included in the systematic review

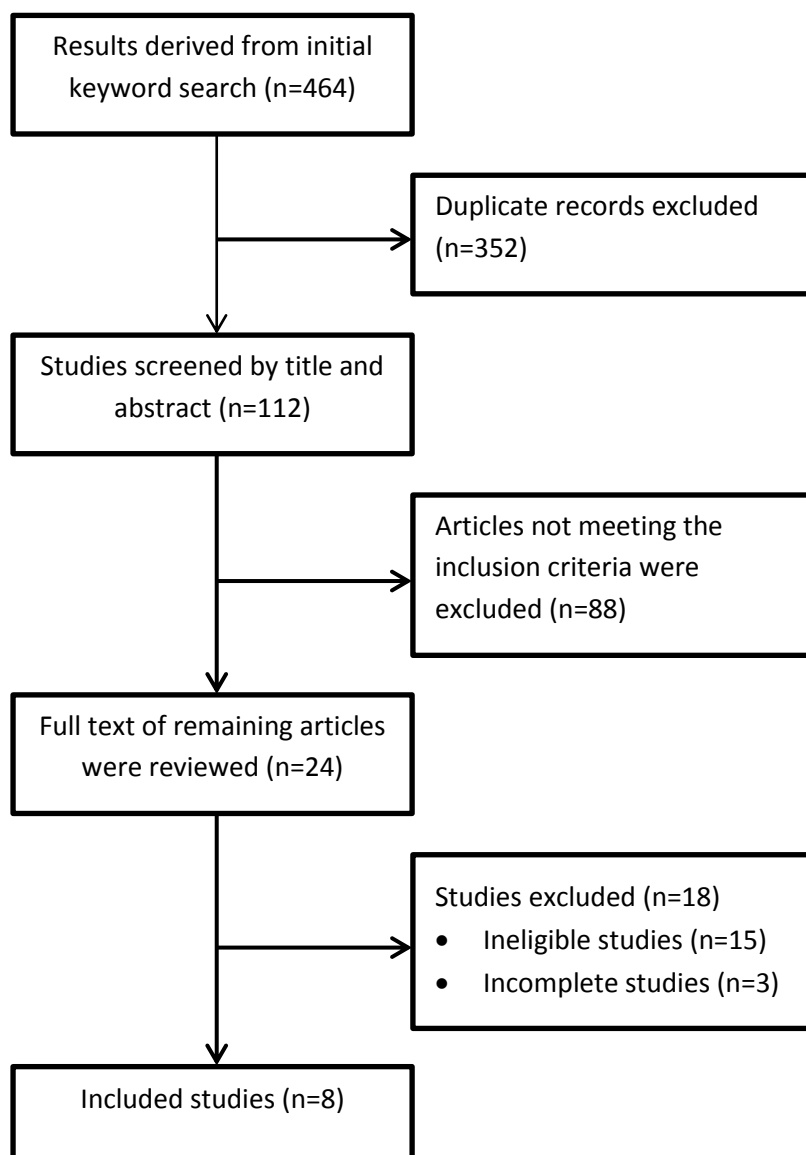**Appendix D:** Summary of outcomes tested

Figure 1: Flow chart showing the retrieval process of studies included in the systematic review

Table 1: Main characteristics of the four stronger studies included in the systematic review

Study ID	Article and study type	n	Population, age range and location	Comparison groups (n)	Concurrent treatment/other materials	Dose of MI	MI training and quality control measures	Participant uptake	Follow up	Outcome
1	Ismail et al (2008), RCT	344	Adults with Type 1 diabetes, age 16-65, UK.	Usual care group (n=121), Usual care with motivational enhancement therapy plus Cognitive Behavioural Therapy (n=106), Usual care with Motivational enhancement therapy (n=117)	None	Patients are offered 4 face-to-face 50 minute sessions	6 months training in MI with supervision. MI Treatment Integrity code used to evaluate a random sample of 20 tapes from each group	66%	3,6,9, & 12 months	Differences between groups in HbA1c levels and other outcome measures (e.g. diabetes self care scores and BMI) were not significant.
2	Smith West et al (2007), RCT	217	Overweight women with Type 2 diabetes (38% African American), mean age: 54, USA.	Motivational interviewing (109), Attention placebo (108)	42-session group weight management programme, average group size was 14 people.	5 additional 45 minute individual MI sessions at 3 month intervals.	Ongoing clinical supervision of MI and intervention protocol fidelity monitoring. Randomly selected audiotapes were reviewed weekly using standardised coding format.	93%	6, 12 & 18 months	MI group weighed significantly less than attention placebo at 6 months (≤ 0.01) and 12 months (≤ 0.02) 18 months (≤ 0.04). Significantly greater A1C with MI group was noted at 6 months (P=0.002).
3	Hokanson et al (2006), RCT	114	Adults with Type 2 diabetes, aged 21-80, USA	Control group (n=57) received MI, Intervention group (n=57) received individual smoking cessation – standard care.	Both groups received identical diabetes education and treatment recommendations for achieving glycaemic control. Nicotine replacement therapy was also offered	1 face to face 20-30 minute smoking specific MI meeting and 3-6 telephone MI sessions	MI provided by researchers who have received 12 hours of smoking cessation training and MI.	60%	3 & 6 months	Intervention groups indicated fewer who smoked daily at 3 month follow up (p= 0.48), but no significant difference at 6 month follow up. No significant differences in A1C, plasma lipids, blood pressure or weight loss.
4	Brug et al. (2007), RCT	209	Newly diagnosed Type	Intervention group: patients of dieticians trained in	Usual treatment	4-5 MI sessions – 1st session	Training was 2 day basic MI skills training	60%	none	A significant reduction in saturated fat intake was found in MI group compared

			2 diabetes patients, mean age: 59, Netherlands	Motivational interviewing (83), Control group is dieticians patients not trained in Motivational Interviewing (59)		was 45 mins, follow up session was approx 15 mins	specifically for dieticians. MITI and MISC were used to evaluate 15-minute transcripts of sessions			to control group (p=0.00). No significant difference was found in fruit or vegetable intake, BMI, waist circumference or HbA1C.

Table 2: Main characteristics of the weaker studies included in the systematic review

Study ID	Article and study type	n	Population, age range and location	Comparison groups (n)	Concurrent treatment/other materials	Dose of MI	MI training and quality control measures	Participant uptake	Follow up	Outcome
5	Smith et al. (1997), RCT	16	Women who 120-200% above ideal weight with Type 2 diabetes, aged >50 year, USA	Standard behavioural weight-control program (10), behavioural weight control program with Motivational Interviewing (6)	16-session group behavioural weight control program	Three sessions, length unknown	MI conducted by psychologists experienced in MI.	72%	4 months	MI group displayed significantly better glucose control than standard behavioural group (p=0.05). Both groups lost a significant amount of weight but differences between groups were not significant. No significant differences between groups in calorie intake or exercise uptake.
6	Calhoun et al (2010), cohort (one group pre-post) study	26	Northern Plains Indians with Type 2 diabetes, age: $\leq$ 18, USA	No comparison group: Participant data was taken 6 months prior to baseline, for the baseline and 3 months after intervention.		2 MI sessions – 30 minutes within 3 weeks of baseline	Interventionist received supervision and attended training in MI skill coding to strengthen MI knowledge	22%	3 months	There was a reduction in reported unhealthy dietary intake, although not significant (p=<0.08), no significant change in glucose was found. Exercise/physical activity was measured but not recorded.
7	Rubak et al (2009), RCT	265	Adults with Type 2 diabetes, age not specified, Denmark	65 GPs were randomised into Control and intervention group. Control group had 128 patients and intervention group had 137 patients, each patient received individual sessions.	GPs for both groups attended a course on intensive treatment for type 2 diabetes.	Unknown.	GPs in Intervention group received 1 ½ day training with a ½ day follow-up twice during 1st year.	87%	12 months	No significant differences in alcohol consumption or smoking between groups
8	Britt et al (2008), experimental.	18	Adults aged 16-69 years, with Type 1 or 2 diabetes, New Zealand	Intervention group: Individual Motivational Enhancement Therapy (MET) (n=9). Control group (n=9)	Patient education	MET is a brief 4 session form of MI by diabetes nurse educators	Intervention was delivered by Nurse educators who were trained in MI	unknown	3 & 6 months	4 of Experimental group and 3 of Control group had clinically significant decrease in HbA1c post intervention and at 6 month follow-up for control group. 5 of the experimental group showed clinically significant decreases at 3 and 6 months follow up. Control group showed little difference in lipids,

								whereas experimental group showed total and ldl cholesterol at 3, 6 and 12 month follow up. -not statistically significant

Table 3: Summary of outcomes tested

Study ID	Article	Exercise/ physical activity	Dietary intake	weight loss	Smoking	Alcohol	HbA1C (Blood Glucose)	Cholesterol	Blood Pressure
1	Ismail et al (2008)	NS	NS	NS		NS	NS	NS	NS
2	Smith West et al (2007)		Sig.	Sig.			Sig.		
3	Hokanson et al (2006)			NS	Sig.		NS	NS	NS
4	Brug et al. (2007)		Sig.	NS			NS		
5	Smith et al. (1997)	NS	NS	NS			Sig.		
6	Calhoun et al (2010)	NS	NS				NS		
7	Rubak et al (2009)				NS	NS			
8	Britt et al (2008)						NS	NS	

NS= MI not significantly effective

Sig.= MI significantly effective