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**THESIS SUBMITTED IN FULFILMENT OF THE DEGREE OF DOCTOR OF
CLINICAL PSYCHOLOGY**

**CITY UNIVERSITY,
LONDON**

DECEMBER 2005

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ACKNOWLEDGEMENTS

I would like to thank my supervisor Professor Dermot Bowler, for his help and encouragement during the whole length of time this thesis has been in preparation. I particularly appreciate his extreme patience and understanding of the challenges of preparing a thesis whilst working full-time in the National Health Service. His comments have always been positive, encouraging and helpful and his tolerance remarkable, even when I have struggled to grasp the whole picture.

I would also like to thank Dr Elizabeth Isaacs for her encouragement and helpful advice particularly during the time when I was submitting the proposal to the hospital ethics committee, a rather long and arduous process. My meetings with her have always been constructive and inspiring.

Thank you to Dr Malcolm Cross who has read different case studies and my literature review with patience and offered very helpful advice about structure and content and inspired me to continue to revise and improve these sections of the thesis.

Nicola Jolleff, Lead Clinician of the Neurodisability Service, Wolfson Centre, Great Ormond Street Hospital for Children, friend and colleague, has been very active and positive in her support, encouraging me and enabling time to collect my data and to write up the results even when clinics have been under pressure of long waiting lists and in need of clinical input from psychology. Choosing between clinical obligations and research interests is a relentless conflict and Nicola has trusted me to achieve some balance in my professional and my personal obligations, for which I will always remain grateful.

All my colleagues at the hospital, including Professor Robert Surtees, have remained patient and supportive throughout and have never seemed to doubt that one day this piece of work will be completed.

Dr Naomi Dale, Consultant Clinical Psychologist, has been positive and encouraging, supporting opportunities to take blocks of study leave when necessary.

I am grateful to my husband, David Brennan, who has accepted without complaint the many weekends I have spent in front of the computer. Also particularly thank you to Charlotte and Jonathan who have grown up into delightful young adults without much input from their mother, who has been preoccupied with fulfilling a personal ambition.

My Australian family, including my mother who is now 89, my brothers Kevin and Phillip and their families and my dear sister Wendy, all remained supportive and encouraging and have waited patiently and continued to remember my preoccupation. I am very grateful for their continuing interest and inclusion of my needs in their life when I am so distant.

Lastly I want to acknowledge the continuing interests and support of my friends and colleagues including Vicky Bianco, Linda McCann, Carolyn Smith and Diane Melvin who have cheered my soul, have stayed positive and kept me going with interest and humour.

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SECTION A

PREFACE

My professional career has focused on the development and emotional well being of children. As a clinical psychologist I have worked in child mental health and in Neurodisability. The present piece of work has arisen out my interest in the links between both areas of work, particularly the links between childhood disability and social competency. When I was training I became particularly interested in a group of children with mild motor difficulties who also had difficulty with social relationships and behaviour. Together with an occupational therapist I set up a group for four young children with the aim of providing practical intervention on motor functioning and visuo-motor skills and to provide some input on behavioural management. This group of children, I now realise, would fit with the children I have investigated in the research presented as part of this thesis.

Over the years I continued to work with children referred to child and mental health but spent several years as a clinical research psychologist investigating behaviour problems in children with Downs Syndrome and children with other learning disabilities (Gath and Gumley 1986, 1986, 1987). I also began working with children and families in a paediatric setting. During this period I was responsible for psychological intervention with children who presented with behavioural or learning difficulties either to the Department of Child and Family Consultation, to Paediatric Outpatient Clinics, or who were identified when inpatients on the paediatric wards. I worked as a member of a multi-disciplinary team of professionals providing a clinical service both to the children and their families. A routine part of my duties in this setting was the provision of psychometric assessments of children referred to the department for an opinion on specific learning difficulties, or questions related to a chronic condition such as epilepsy, investigating links between cognitive functioning and behaviour at home or at school. My interest in the cognitive and

emotional development of children with chronic conditions and disability developed during this period. I became aware of the impact, offering support and advice to families with concerns about their child's behaviour and learning needs could have on future progress. This can involve establishing links with schools and providing information and advice when the child's behaviour or medical condition caused particular concerns in the educational setting. During this period I was also working in the Neurodevelopmental Assessment Clinic, which was part of the Neurodisability Service at Great Ormond Street Hospital for Children. I supplied the psychological input to the multidisciplinary team. I also provided specialist tertiary level consultation on diagnosis and management of children with complex Neuro-developmental problems where an additional opinion had been requested by secondary level services. The children had a range of multiple disabilities, including physical and visual disabilities. These assessments included neuropsychological assessments as well as assessment and advice on complex behavioural difficulties seen in children with complex learning disabilities. I advised on educational placement and rehabilitation needs and where local services did not have the expertise, I provided brief intervention in the form of advice on behaviour management particularly in the context of specific neurodevelopmental disabilities.

I began the main research project when working in paediatric, mental health and Neurodisability settings. I was struck by the similarities between a group of children referred to each service, particularly those with mild motor impairment in combination with difficult behaviour at home or at school. The children were usually over six years and presented with very unusual behavioural difficulties in association with learning problems. The cognitive profiles of these children appeared to fit that described by Rourke and colleagues (1989) as Non-verbal Learning Disorder. However I also noted the overlap

between the description of learning and behavioural difficulties described as a feature of this condition with the patterns of behaviour described as Asperger Syndrome. Although initially interested in investigating the group described by Rourke and colleagues, access to an appropriate study group was not possible and the present research developed out of my interest in the links between a group of children with a diagnosis of Developmental Coordination Disorder and those children with a diagnosis of Autistic Spectrum Disorder (ASD) such as Asperger syndrome

Around that time I changed jobs and began to work with the Neuro-oncology team in Great Ormond Street Hospital. I had responsibility for re-establishing a service providing psychological input to the Neuro-oncology service. I provided advice on behavioural and emotional difficulties, educational placement and special needs of children who have had treatment for brain tumours. Among other duties, this involved carrying out psychological assessments of children with complex difficulties. I developed a database to record relevant information on cognitive and behavioural profiles of the children and have developed research programmes specifically to look at long-term effects of treatment on cognitive, emotional and behavioural outcome.

I have noted and become interested in the social competencies of those children who have acquired disability, including motor disability, through diagnosis and treatment for brain tumours and those who have developmental difficulties such as the group of children investigated in this research. The literature has begun to describe the pattern of cognitive functioning seen in some children following treatment for brain tumours as being best described as non-verbal learning disorder.

The theme running through this thesis is the relationship between 'normal' IQ and more subtle difficulties with learning and social interaction, which can have a significant disabling

effect on the child's functional skills, their ability to relate to their peers and their ability to learn in an educational setting.

Section B is an investigation into social and behavioural profiles shown by a group of children with Developmental Coordination Disorder (DCD), with the aim of providing clarification of patterns of difficulties.

The case description in Section C provides an example of a child who has been diagnosed and treated for a brain tumour, who, whilst apparently well and with good prognosis, continues to have significant effects several years following treatment. The interplay between her emotional well being, late effects and her ability to function with her peers is having an ongoing impact on her capacity to return to a 'normal' life. Encouraging an appropriate level of support from the educational services is a continuing concern as her difficulties are subtle and not immediately apparent to her teachers or her peers. Similarly many children who were part of the main study had difficulties relating with their peers, and were not receiving additional support in school unless their pattern of difficulties were obvious and causing disruption in the classroom. The case study demonstrates the way I aim to work as a clinical psychologist, developing links between different features of a child's learning profile and emotional status and providing proactive intervention both with the family and with local services.

The review of the literature in Section D aims to investigate the immediate and long-term effects on children of diagnosis and treatment for brain tumours. The review confirms that the difficulties experienced by the child described in Section C are common, and highlights the need for further investigation into the development of appropriate rehabilitation interventions. With recent developments in medical treatment more children are surviving throughout childhood and into adult life. Some children are surviving with significant

disability but others, although often coping well, have difficulties in all areas of functioning, which have an impact on their emotional well being and their ability to live an independent adult life.

The thesis therefore aims to bring together the different aspects of cognitive, behavioural and social concerns that will impact on the lives of children with developmental difficulties and those with acquired long term chronic illness related impairments, resulting from the effects of diagnosis and treatment for brain tumours.

Gath A & Gumley D., (1986). Behaviour Problems in Retarded Children with Special Reference to Down's syndrome. *British Journal of Psychiatry* 149 pp.156-161

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SECTION B

BEHAVIOUR AND SOCIAL FUNCTIONING IN CHILDREN WITH DEVELOPMENTAL COORDINATION DISORDER

Submitted in part fulfilment of the degree of D. Psych of City University, London

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ABSTRACT

The present study investigated the behavioural profile shown by a group of children with Developmental Coordination Disorder (DCD), to provide clarification of patterns of difficulties, and to examine how they relate to the diagnostic criteria of an Autistic Spectrum Disorder. Children aged between 6 and 12 years were recruited to the study from three sources: a neurodevelopmental assessment clinic, the Dyspraxia Foundation and through informal contact. Informed consent was given by 107 families. Parents completed the Movement Assessment Battery Checklist, (Henderson & Sugden 1992) the Rutter A (2) parent questionnaire (Rutter, Tizard & Whitmore 1970), and an adapted version of the ASSQ (Ehlers & Gillberg 1993). Children with reported motor difficulties were found to have significantly more clinical behavioural difficulties as measured by the Rutter A (2) Scales. They also had significantly higher scores on the adapted ASSQ (AASSQ) than those without motor difficulties. Results indicate a significant positive correlation between degree of motor difficulty and scores on both the behaviour questionnaire and the AASSQ. The implications for clinical management and educational support are discussed.

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IN

ORIGINAL

CHAPTER 1

INTRODUCTION

1.1 Aim

The present study aimed to investigate the behavioural profile shown by a group of children with Developmental Coordination Disorder (DCD), to provide clarification of patterns of difficulties, and to examine how they relate to the diagnostic criteria of an Autistic Spectrum Disorder (ASD), such as Asperger Syndrome.

1.2 Background

My clinical work highlighted a series of children referred to a Neurodevelopmental Assessment Clinic, for consideration of learning difficulties and a diagnosis of Dyspraxia or Developmental Coordination Disorder (DCD). These children were noted to have motor difficulties, and appeared to perform significantly worse than their peers on motor tasks. Their parents also described difficulties with peer relationships and social interaction. Clumsiness, or difficulty with motor skills, is commonly seen in children with an Autistic Spectrum Diagnosis, such as Asperger syndrome (Wing 1981; Gillberg 1989; Szatmari 1989) and together with social and behavioural difficulties can be one of the first characteristics that cause concern for parents (Howlin & Moore, 1997). There is a likelihood that a significant number of children with a diagnosis of DCD would also fulfil criteria for an ASD diagnosis and the present study aims to investigate this association.

1.3 Developmental Coordination Disorder (DCD)

Both ICD 10 (WHO, 1990) and DSM IV (APA, 1994) have provided an account of the features characteristic of this pattern of developmental difficulties and Developmental Coordination Disorder (DCD) is now the term most commonly used both in the literature and in clinical practice to describe children with motor impairment.

1.3.1 Diagnostic Criteria – Developmental Coordination Disorder

The ICD 10 classification for Developmental Coordination Disorder (DCD) or Specific Developmental Disorder of Motor Function (SDD-MF) requires the child's score on a standardised test of fine or gross motor co-ordination to be at least two standard deviations below the level expected for the child's chronological age. It is a disturbance in childhood that significantly interferes with academic achievement or with activities of daily living. It is usual for the impairment to be associated with weak performance on visuo-spatial tasks. There is no diagnosable neurological disorder, acquired or congenital, and manifestations of the child's motor difficulties may vary with the child's age and development. The disorder is not solely explained by general intellectual impairment. It can be diagnosed as part of a mixed specific developmental disorder, in association with speech and language disorders, and difficulties with literacy or numeracy when there is a major difficulty over all of these disorders. The ICD-10 classification specifically mentions that there may be associated socio-emotional behavioural problems although this is not a diagnostic criterion.

The DSM 1V (American Psychiatric Association, 1994) classification describes Developmental Co-ordination Disorder as a marked impairment in the development of motor co-ordination, which is not due to a general medical condition. The degree of motor impairment in a child can vary according to the child's age and developmental level or cognitive skills. The disturbance significantly interferes with academic achievement or activities of daily living. The inclusion of DCD in the DSM IV manual suggests an association with behavioural disorders but they are not a specific requirement of the diagnosis and there is an explicit statement that the disturbance does not meet criteria for Pervasive Developmental Disorder. Pervasive Developmental Disorder is described as a

disorder that primarily affects verbal and non-verbal communication, social skills and imaginative play and is often seen in association with significant learning disability. Developmental Coordination Disorder as defined by ICD 10 and DSM IV is now the most common term used to describe children with motor difficulties. However, other terms have been used in the literature to depict the characteristics of these children, including clumsiness (Ayres 1972; Gubbay 1975; Henderson, 1987), developmental dyspraxia, (Cermak, 1985; Dewey, 1995) minimal brain dysfunction, (Rutter, 1982) and neuromotor soft signs (Shaffer, 1985).

Clumsiness has been used to refer to the child whose ability to perform skilled movements is impaired, despite normal intelligence and normal findings on neurological examination (Gubbay, 1975). It has also been used to describe a disorder of sensory integration, which interferes with the child's ability to plan and execute skilled and non-habitual motor tasks (Ayres, 1972).

Developmental Dyspraxia has been used to refer to children demonstrating motor planning difficulties, whilst the term 'minimal brain dysfunction' describes a group of children with motor impairment who lack both a history of brain damage and abnormal neurological signs (Rutter, 1982). Rutter (1982) suggests the term may have originated in the 1920's when hyperactivity, antisocial behaviour and emotional instability began to be seen as an indication of brain damage. 'Soft signs' of neurological impairment usually refer to perceptual-motor dysfunction, abnormal movements and motor co-ordination problems. Different labels have therefore been used to describe a developmental problem in children who have never developed normal motor skills. There can be accompanying soft neurological signs such as motor awkwardness, and awkward gait. More recently the

literature has indicated a link between clumsiness or DCD and neonatal problems (Gubbay, 1978; Holsti, Grunau & Whitfield, 2002).

1.3.2 Clinical Features of DCD

Despite the range of words used to describe the condition there is some agreement on the main features, which are commonly seen in the children.

- **Motor skills**

Children usually show developmental delays in sitting up, crawling, and walking. They may show deficits in handwriting, problems in gross motor coordination (jumping, hopping and standing on one foot) and problems in fine motor coordination (tying shoelaces, tapping one finger to another). They are also reported to be slow to learn activities of daily living such as dressing skills, feeding independence including handling a knife and fork, and personal grooming.

- **Behaviour Emotional and Social Development**

Associated behavioural and social difficulties in children with DCD have been documented in a number of studies (Cermak 1985; Henderson 1989; Losse 1991; Schoemaker & Kalverboer 1994) and have typically been attributed to the child's strategies for coping with their motor difficulties (Gubbay, 1975). Children have been described as showing negative behaviour, low self-esteem and difficulties with peer relationships (Gubbay, 1975; Ayres, 1980; Cermak, 1985).

Parents of children with DCD describe patterns of behaviour, which are largely attributed to the difficulties of coping with the child's motor problems. They describe poor visual and auditory memory, with resulting difficulty in remembering possessions. The children have problems with absorbing and recalling information, immature social skills, literal use of language, disruptive behaviour, impulsive behaviour, and difficulty controlling emotions.

They are often described as loners, who have problems forming relationships and appear isolated from their peers (Dyspraxia News, 1997).

It has been suggested in the literature that social and behavioural problems are a consequence of the impact of the motor difficulties on the child's emotion and behaviour. The problems were seen as a direct consequence of the value placed on physical skills by the child, the family and society in general (Gubbay, 1975).

- **Core features**

In summary, although there has been a different emphasis on the common features of children with motor difficulties, the literature highlights four core features, which remain common to all descriptions of these difficulties whichever label is used. These include impaired motor performance, absence of neurological signs, normal intelligence, and discrepancies in motor capacities. The other features commonly described, including difficulties with literacy and numerical skills, speech defects, over activity and distractibility, and emotional or behavioural difficulties, are usually attributed to low self-esteem (Gubbay, 1975; Ayres, 1980). Throughout the literature it has been emphasised that it is important to recognise the nature of the child's motor difficulty and give treatment, because the children have a high risk of developing emotional and behavioural disorders (Gubbay, 1978, 1979; Cermak, 1985).

1.3.3 Motor Impairment in children with Developmental Coordination Disorder

Although both the DSM-IV and ICD-10 have attempted to formalise the criteria needed for a diagnosis of DCD, there has been little agreement in the literature on the diagnostic criteria required to fulfil the criterion for motor impairment. Neither DSM-VI nor ICD-10 criterion provide guidelines on what specific type, if any, of motor impairment is

characteristic of this condition. For example, no distinction has been made between fine and gross motor difficulties, whether both are a necessary feature or if difficulties with one area would be sufficient to fulfil the criteria for a diagnosis. As a result there have been various studies of different aspects of motor impairment including the studies by Dewey (1995) who emphasizes that developmental Dyspraxia, another term used to describe children with difficulties similar to DCD, is a disorder of gesture. Studies have investigated specific motor skills, such as finger tapping, ball skills, self-help skills or gross motor skills such as riding a bike. Assessments of these various skills in the literature are not always related to age or intellectual function, whereas the DSM-IV and ICD-10 classifications emphasise the relationship of motor impairment to the child's age and intellectual level. Other authors (Missiuna & Polatajko, 1995) have queried whether the diagnostic category of Developmental Coordination Disorder includes children with developmental Dyspraxia, or children with sensory integrative dysfunction.

However since the publication of the ICD-10 and DSM-IV criteria for DCD, there has been more agreement in the literature about the level of impairment, although not necessarily on the type of motor impairment, that has been measured, and more studies are reporting results of standardised tests.

1.3.4 Prevalence Rates of Developmental Coordination Disorder

From a survey of 992 children from five mainstream schools Gubbay identified 56 clumsy children (Gubbay, 1975). He concluded that this indicated approximately 5% of 8 – 12 year old children have motor impairments. Others have suggested 10% (Henderson & Sugden, 1992). Kadesjo and Gillberg (1999) looked at prevalence rates in a total population sample of 7 year olds and found 4.9% had severe DCD and 8.6% had moderate DCD. Kadesjo and Gillberg found the rate among boys in the population were 8.0% and among girls was

1.1%, although Gubbay in his 1975 study reported almost equal incidence of males and females. The range of terms used to describe the condition may have meant different diagnostic criteria have been used, but overall there seems to be general agreement that that between 5% and 10% of children have difficulties with motor coordination with higher prevalence rates among boys than girls.

1.3.5 Prevalence rates of disturbed behaviour in children with Developmental Coordination Disorder

There have been few attempts in the literature to provide a specific description of behaviours, or to investigate the links between behavioural difficulties and motor impairment. As a result the terms behaviour or behaviour problems when used to describe associated behavioural difficulties in children with motor impairment, tend to be imprecise and wide ranging.

The literature on children with Developmental Co-ordination Disorder frequently refers to existence of disturbed behaviour, including impairments in attention (Gillberg & Gillberg, 1989), poor self-esteem and motivation (Shoemaker & Kalverboer, 1994), social immaturity, low self-esteem, anxiety, and disruptive behaviour (Henderson & Hall, 1982; Henderson, 1992). Studies of developmental dyspraxia have also supported the view that it involves a linguistic conceptual disturbance, is a disorder of gesture (Dewey, 1995) and that there is a high incidence of neurobehavioural difficulties (Deuel & Doar, 1992).

However, although several studies have investigated specific aspects of behavioural or emotional functioning such as attention, hyperactivity, anxiety or depression, few have aimed to describe an overall or global picture of the child's social emotional and behavioural functioning. To avoid confusion the term 'behavioural difficulties' will be used in the present study as a global term to describe all aspects of behavioural and emotional

functioning reported by the literature, particularly when the reported behaviour is not described in detail.

There have also been few studies, which have reported on the prevalence rate of associated behavioural difficulties in DCD. Bax and Whitmore (1987) followed a group of children who had a neurodevelopmental assessment at five years which was then compared with the children's educational and behavioural outcomes at the ages of seven and ten years. They carried out examinations of three hundred and fifty one children on entry to primary school. The examinations included an interview with parents, administration of the Rutter A behaviour scale, tests of hearing and vision and a full paediatric examination, including fine and gross motor function. Children with poor motor skills, measured during the paediatric examination, showed a significantly higher rate of learning difficulties at age ten than children with normal motor scores. Of a total of thirty children who had motor scores above the mean for the whole group, that is significant motor difficulties, but normal ability scores at age five, seven had learning or behavioural difficulty at ages seven and ten.

Gillberg and Gillberg (1989) reported that 65% of children with motor perceptual problems had attention deficit disorder. More recently Kadesjo and Gillberg (1999) carried out a population study of 818 seven year olds and found approximately half of all children with DCD had moderate to severe symptoms of attention deficit/hyperactivity disorder (ADHD). Kadesjo and Gillberg (1999) reported that four children in the DCD group met full Gillberg criteria for Asperger syndrome (Gillberg, 1991), and they also found symptoms of Asperger syndrome were predicted independently by both DCD and ADHD. The children with DCD had high rates of Oppositional Defiant Disorder but it is not clear what percentage of the DCD group, either they or the Asperger group represents (Kadesjo & Gillberg, 1999). Because these studies focused on the presence of ADHD they do not

provide an overall rate of all behavioural difficulties, which may be seen in children with motor impairment.

1.3.6 Behaviour and emotional disturbance in children with Developmental Coordination Disorder

In the literature on children with motor impairment, where the aim was to look specifically at motor skills, there are frequent references to behavioural difficulties seen in the children even when children with more obvious difficulties with behaviour have been excluded from the study group (e.g. Dewey & Kaplan, 1994).

A study by Dewey and Kaplan (1994) to investigate whether subtypes of developmental motor deficits could be identified looked at 102 children aged 6-10 years; 52 were identified as displaying deficits in motor skills and 51 were typical controls. Cluster analyses indicated four sub-groups of children. One group had deficits in motor sequencing; another had deficits in motor balancing, coordination and gesture, a third had severe deficits in all areas and the fourth had no motor deficits compared with other groups. They excluded children who had evidence of attention deficit hyperactivity disorder or who were taking medication for mood or behavioural disturbances. Dewey and Kaplan (1994) reported that among children with developmental motor deficits, those who showed the greatest deficits in motor skills were also the most impaired on a test of receptive language skills and that deficits in motor sequencing and language comprehension appear to co-exist. Dewey indicated children with more severe dyspraxia or DCD are at increasing risk of behavioural difficulties.

A further investigation by Dewey and colleagues (Dewey, Kaplan, Crawford & Wilson 2002) investigated problems of attention, learning and psychosocial adjustment seen in children with DCD and concluded that all children with DCD are at risk for problems in

these three areas and suggested that these problems should be investigated in all children regardless of the degree or severity of associated movement problems.

Whilst some studies (Dewey & Kaplan, 1994) that look specifically at motor skills exclude children who are reported to show behavioural difficulties, others, (e.g. Henderson & Hall, 1982) although containing references to the presence of behavioural difficulties, have not used formal measures to identify these difficulties nor has the incidence of behavioural difficulties in the study population been reported.

In an investigation of children referred to a medical facility because of school problems, Deul and Doar (1992) found a high incidence of Dyspraxia, which they defined as an inability to learn or perform serial voluntary movements with proficiency expected for age or verbal intelligence. These children were described as socially immature and some had begun to develop more negative behaviour and attitudes to school than academically average children, although it is not clear how the authors measured the behavioural difficulties described in these children.

A later study by Schoemaker and Kalverboer (1994) aimed to examine patterns of co-occurrence of social and affective problems and to assess their association with the severity of movement disorder. The children were screened for motor problems by school doctors and were then assessed on the Henderson revision of the Test of Motor Impairment (Stott, Moyes & Henderson, 1984). The 5% boundary of the lowest performing children was used as the criterion for selecting children to the study. Eighteen children with a mean age of 7 years 4 months were included in the study. A control group of children without motor problems was also selected. Teachers and parents completed questionnaires to measure social behaviour. Teachers reported that the children differed from the controls on measures of introspection such as being more serious, more insecure and more isolated.

Parents reported that children differed from controls on extroversion and introversion but not in any other way. Parents also reported that 50% of the motor impaired group had difficulties making contact with other children. Parents of children in the control group did not report these difficulties. Schoemaker and Kalverboer (1994) concluded that clumsiness is not an isolated problem and that it is associated with various social and affective problems even in young children. They did not find a relationship between severity of clumsiness and affective problems except for 'socially negative behaviour'. They found signs of socially negative behaviour were less common in the most severely affected children. Interestingly, they also mentioned that individual data revealed that almost all the children who were clumsy had social or affective problems irrespective of the degree of their motor difficulty.

A study by Soorani-Lunsing (1993) explored the specific relationships existing between behavioural or cognitive problems and certain types of minor neurological dysfunction, in a group of twelve-year-old children of whom 172 had minor neurological dysfunction and 174 were typically developing children. The behaviour was evaluated by asking parents to complete the Children's Behaviour Checklist (Achenbach, 1982) and the Rutter Teachers Scale (Rutter, Tizard & Whitmore, 1970). The presence of fine manipulative disability contributed to nearly all behavioural and cognitive problems. Co-ordination difficulties were related to school failure. Hypotonia was related to fearfulness and clumsiness at home and school, and to other behavioural difficulties at home. They also found a relationship between fine manipulative difficulties, hypotonia and difficulties making social contacts.

Neurological soft signs were also found to be markers for cognitive and psychiatric problems as measured by parent and teacher checklists in low birth weight children in a study by Breslau and colleagues (2000). Children born prematurely, who have extensive

perceptual or motor problems, have also been shown to be more likely to have problems with cognition, reading and behaviour (Jongmans, Mercuri, Dubowitz & Henderson, 1998). Studies by Szatmari and Taylor (1984), and Taylor, Powell, Cherland & Vaughan, (1988), found that children aged 7 - 10 years who display high levels of abnormal motor movements also manifested behaviour problems at school and were judged to perform poorly at school. They concluded that boys who show abnormal movements seemed to display more cognitive and motor problems than do girls who demonstrate abnormal movements.

The literature has begun to show an association between motor impairment and behaviour problems both at home and at school. These problems are variously described as including difficulty with attention, social and affective problems, social immaturity and negative social behaviour.

1.3.7 Prognosis /long-term outcome in children with DCD

There has been a growing literature indicating that neuromotor soft signs have prognostic value for later behavioural, cognitive and academic functioning. The literature has indicated that 'soft signs' of neurological impairment (e.g. perceptual-motor dysfunction, abnormal movements, motor co-ordination problems) seen in some children may have considerable prognostic value for later behavioural, cognitive and academic functioning (Carte, Nigg & Hinshaw 1996; Landgren, Pettersson, Kjellman & Gillberg, 1996; Raine Brennan, Mednick & Mednick 1996). While soft signs may suggest problems in neurological functioning, by definition they cannot be linked to 'hard' (i.e. localised) signs of neurological abnormality and they do not constitute a diagnosable neurological condition such as cerebral palsy. Shaffer and colleagues carried out one of the earliest studies (Shaffer et al 1985). They found a relationship between neurological soft signs, at age seven, and subsequent anxiety

disorders at age seventeen in a birth cohort of 63 boys and 26 girls. Poor co-ordination signs were found to be over-represented in the anxiety-withdrawal group at age seven and 81% of children with an anxiety-withdrawal diagnosis at age seventeen had had a sign reflecting a disorder of co-ordination at age seven. The assessment of anxiety at age seven was based on the child's behaviour during psychological testing at that time. Shaffer et al., (1985) concluded that of the children who had soft signs and who also displayed anxious dependent behaviour during psychological testing at age seven, just under half went on to show anxiety or affective disturbance in later adolescence.

A study of very low birth weight infants by Marlow, Roberts, and Cooke (1993), reported the presence of motor problems at 6 years was the best predictor of school problems at 8 years. In addition to academic performance deficits, they also found a link between neuromotor soft signs and behavioural difficulties such as emotional disorders and over-activity, identified by teachers.

More recent literature on childhood neurological soft signs has also shown that they are associated with concurrent and later problems in academic functioning, cognition, behaviour problems and psychiatric symptoms (Pine, Shaffer, & Schonfield, 1993; Malla, Norman, Aguillar, & Cortese, 1997; Pine, Wasserman, Fried, Parides, & Shaffer, 1997; Foulder-Hughes & Cooke, 2003).

Pine and colleagues (1997) investigated the stability of soft signs and behaviour over a period of one year in a group of 48 boys aged 8.5 years at the first assessment and 9.7 years at second assessment. During both assessments, symptoms of psychiatric disorders based on DSM-111-R guidelines were assessed by standardised parent interview. They investigated ADHD, oppositional defiant disorder, and conduct disorder. They also investigated symptoms of anxiety and depressive disorders. At both assessments the

children had a medical examination where soft signs (motor slowness, accuracy, abnormal movements and smoothness) were rated. Soft signs showed a high degree of stability over one year. Soft signs also showed significant associations with the four psychiatric scales, oppositional defiant disorder (ODD), conduct disorder (CD), anxiety/depression and ADHD. They found a significant association between soft signs and anxiety/ depression at time one and time two, and a significant association between ODD/CD and soft signs at both time one and time two, but no significant association between ADHD and soft signs at either time. Further analyses suggested that ODD/CD were more closely related to soft signs than persistent anxiety and depression. Pine et al., (1997) concluded that motor impairment in young boys is related to stable forms of psychiatric disorder.

A prospective study by Walker, Downey and Bergman (1989) reported on behavioural data collected on 107 children (mean age of 9.7 years) of normal and psychiatrically disturbed parents, at two points in time. At the first assessment the Bruininks-Oseresky Test of Motor Proficiency (Bruininks 1978) was administered to the children and parents completed the Children's Behaviour Checklist (Achenbach, 1982). Parents completed the Children's Behaviour Checklist again for a follow-up assessment one year later. The pattern of results suggested poor motor skills were linked to increased problems in attention and social functioning. Walker et al, (1989) also reported a correlation between motor function, behavioural difficulties and age.

A comprehensive study of clumsiness in school children by Henderson and Hall, (1982) described three groups of children. From a sample of approximately 400 children attending four infant schools, teachers identified twenty children on the basis of two sets of criteria: poor motor coordination for age, and lack of coordination which was significantly affecting school progress. The children were matched with controls and each child was given four

tests: a neurodevelopmental examination, the Motor Impairment Test (Stott, Moyes & Henderson, 1984), the Wechsler Intelligence Scale for Children (Wechsler 1992) and the Schonell Reading Test (1952). As part of the neurological examination, the child's speech and language skills and social competence were assessed through discussion with the parent. There does not appear to have been a standardized assessment of the child's social competence or other behaviour. As a result of these investigations, three groups are described. One group had motor impairment associated with numerous other problems, including IQ in the lower range, low academic attainment, and some had required speech therapy. This group of children were also described as socially immature and were reported to have behavioural difficulties, although how these manifested is not reported. There was evidence of developing negative attitudes to school. Another group of children were of above average intelligence, and their teachers considered them competent in both reading and number. Their motor impairment seemed to be an isolated problem although the authors mention minor difficulties in social integration in two of these children. The third group of children were of mixed ability, and showed a wide range of scores on both a motor impairment test and neurodevelopmental battery. They were not readily classifiable into either of the two preceding groups. The clumsy children had more problems during their pre-school years than their controls and the number of significant medical events recorded was significantly less in the controls.

The results of Henderson's 1982 study suggested an association between social difficulties, speech delay and motor impairment but the authors indicated that the relationship was too variable for any syndrome to be delineated. However, Henderson and colleagues returned ten years later to study the group who were then aged between 15 - 17 years (Losse et al 1991). They assessed the children on five measures: neurodevelopmental status, general

motor competence, intelligence, self-concept and leisure interests. Information was collected from parent reports and school records. At the time of follow-up Losse et al (1991) found from the school records that the 'clumsy' children had more behavioural difficulties than their normal controls. Of the seventeen children in the 'clumsy' group, thirteen had emotional or behavioural difficulties such as lack of confidence, shyness, difficulty forming relationships, and were described as immature, lonely, and isolated. Seven of this group were also described as having either poor or very poor social self-concept. The results of this study indicate that the emotional and behavioural difficulties described are an important facet of the development of children with DCD and that they have implications for the child's social development.

A series of studies, which investigated the continuing presence of motor difficulties or DCD in children, have also looked specifically at the behaviour seen in these children at time of follow-up, although behaviour has not been specifically investigated in the initial study. As with the Henderson studies the associated difficulties with behaviour appear to have become more apparent as the children have grown older. Cantrell, Smyth and Ahonen, (1994) reported on a follow-up at age fifteen, of a group of children with motor impairment who were first seen at the age five. The children were selected from a population of five years olds in one Finnish town and were first followed-up at the age of eleven years (Ahonen, 1990). At the age of eleven, half of these children still had problems with motor control and were reported by teacher assessment using the Conner's teacher's scales (Goyette, Conners & Ulrich, 1978) to have more problems with inattention and passivity than the control children. On a parent questionnaire (Achenbach, 1982) they were described as childish and immature, with low frustration tolerance and difficulties with peer interaction. At the age of fifteen they were re-assessed on an experimental version of the

Movement Assessment Battery for Children (Henderson, Sugden, Geuze & Losse, 1990). Their self-image was investigated using the Self-perception Scale for Adolescents (Harter, 1989) and a semi-structured interview. The study found that 46% of children who were identified at age five continued to have motor problems at age fifteen. That group was also found to be less sociable than another group who had less extreme problems with motor impairment at age fifteen. Cantrell and colleagues (1994) concluded that the social and educational outcomes are poorest for those with the most extreme motor difficulties at five years old and for those with motor problems associated with lower intellectual skills.

1.3.8 Developmental Coordination Disorder and Asperger Syndrome

Gillberg and colleagues (1982, 1983, 1989), investigated children with problems in motor control. Gillberg selected 59 children who had problems with motor control and symptoms of attention deficit. As a result of these studies a group of children were identified whom he diagnosed as suffering from DAMP - deficits in attention, motor control and perception. Although children showing other behaviour or emotional difficulties were not selected in these studies, Gillberg later refers to associated psychiatric problems including depression, conduct disorders and autistic traits. He also mentions anxiety, low self-esteem and psychosomatic problems, and suggests autistic features can be present in at least half of severe cases of DAMP.

As a result of these studies, including a population based study of six year old children in Goteborg (Gillberg, Rasmussen, Carlstrom, Svenson & Waldenstrom 1982), Gillberg and Gillberg (1989b) found 1.2% of the children without learning disability showed a combination of severe motor clumsiness and attention deficit disorder, and another 10% of the population showed motor clumsiness without attention deficit disorder. Two child psychiatrists examined the children and a comparison group of 51 normal children. They

investigated disturbance in the field of social relationships, including social reciprocal interaction, disturbance of speech and language, and stereotyped behaviours. Eight children from the group with severe motor clumsiness and attention deficit disorder showed autistic behaviour and three boys fulfilled the criteria for Asperger syndrome. Gillberg and Gillberg (1989) have suggested that severe DAMP may exist on a continuum of autistic spectrum disorders.

A follow-up study of children with deficits in attention, motor control and perception (DAMP) by Hellgren, Gillberg and Gillberg, (1994) found a high rate of behavioural, emotional and psychiatric problems in children who had both motor control/perception dysfunction and Attention Deficit Disorder diagnosed at age seven. Based on independent psychiatric examination two thirds of these children had marked psychiatric abnormality. Eight children in the severely affected group had autistic traits (Gillberg 1991), and others in the mild to moderately affected group had depressive disorders at age seven. At age ten, 70% of the children had behavioural and emotional problems and at age thirteen 64% of children were reported to have these problems.

The series of studies by Gillberg and colleagues initially concentrated on those children who were showing the combination of problems, both motor control/perception dysfunction and Attention Deficit Disorder diagnosed at age seven. When the children were sixteen years, a Personality Disorder Examination was used to make diagnoses according to DSM-III (APA, 1980) and DSM-III-R (APA, 1987) (Hellgren, Gillberg, Bagenholm & Gillberg, 1994).

Hellgren and colleagues found that at follow-up, at the age of sixteen years, low non-verbal IQ and poor results on the digit symbol subtest on the WISC-R, a diagnosis of autistic traits or anxiety disorder at age seven years, and minor neurological dysfunction,

amongst other factors, all tended to predict relatively poor outcome at age sixteen. At age sixteen a diagnosis of Asperger syndrome was given for 23% (three males) of the children who had severe motor control/perception dysfunction and Attention Deficit Disorder diagnosed at age seven. These three males had had a diagnosis of Asperger syndrome in the original study. No other children were given a diagnosis of Asperger syndrome. Personality disorders, involving social negativism or withdrawal occurred in 41% of the group, compared to 9% of the control group. Hellgren et al (1994) concluded that the finding from this follow-up study agreed with the population based study by Schaffer et al (1987), where the authors found that impaired motor coordination at age seven predicted the presence of affective and anxiety disorders at age seventeen. Hellgren et al (1994) also found poor psychiatric outcome tended to be predicted by the presence of motor coordination problems in children with attention deficits.

There is therefore a considerable body of evidence to suggest a link between difficulties with motor coordination and continuing behavioural difficulties in association with difficulties with attention and learning (Skinner, & Piek, 2001). Difficulties with motor coordination problems tend to co-occur with other developmental problems in children and there are indications from the literature that this is usual rather than an exception. Neurological soft signs have also been noted to co-occur with behavioural difficulties in children. However, like the studies on children with DCD, standardized assessments have not always been used to assess the children's behavioural status.

1.3.9 Co-morbidity: Developmental Coordination Disorder and other Developmental Disorders

Children with specific language impairment have been described as presenting with motor coordination difficulties and have been shown to have significant problems with motor and

visuo-perceptual tasks, which are similar to those seen in children with Developmental Coordination Disorder (Hill, Bishop, & Nimmo-Smith, 1998; Powell, Bishop 1992; Bishop, & Norbury 2002). Most of these studies have not investigated the presence or absence of associated difficulties with social interaction although this is a common feature of children with specific language disorder. Bishop and Norbury (2002) found that two out of eight children with specific language impairment scored above the cut-off for autistic disorder on the ADI-R (Lord C., Rutter M. & Le Couteur 1994) but did not comment on the presence of any motor difficulties in this group of children. Owen and McKinlay (1997) found concern about fine or gross motor problems, or clumsiness had been expressed for nine out of sixteen children with developmental speech and language disorders, and concerns about behaviour and emotional difficulties had been expressed about nine of the group of sixteen children, although more detail about these difficulties is not provided.

A study which compared 244 children referred because of learning and attention difficulties (but not motor difficulties) and 155 typically developing children, by Kaplan, Wilson, Dewey, and Crawford (1998) found a high number of children with DCD and a high degree of comorbidity between DCD and other developmental disorders including attention-deficit/hyperactivity, and reading disability. They proposed that the comorbidity seen in childhood developmental disorders reflects a single underlying aetiology, which they term atypical brain development. This study confined itself to looking at two aspects of comorbidity and the authors do not refer to social interaction skills of the children.

1.3.10 Social Interaction and Developmental Coordination Disorder

Although there has been work on the links between DCD or soft motor signs and behavioural and learning difficulties, with descriptions of children with hyperactivity, ADHD, obsessive disorder, internalising behaviour, and depression in adolescence, few

studies have provided a detailed picture of these behaviours or of the child's social interaction.

There has been very little work describing the social interaction skills of children with DCD although the literature has established links between DCD and behavioural difficulties, particularly ADHD. The World Health Organisation (1993) has noted an increased incidence of motor skills disorder co-occurring with attention deficit disorders, and hyperactivity is a common feature of autistic children. Early studies of clumsy children referred for medical attention has mentioned that they are referred because of symptoms of anxiety, (Gordon & McKinlay, 1980) or because they are socially inept (Hall, 1988) but these symptoms have not been extensively investigated.

Many of the studies, which do look at behavioural difficulties in children with DCD, Dyspraxia or soft neurological signs, describe children who have emotional difficulties such as anxiety, and difficulties with forming peer relationships, and who are often described as isolated (Schoemaker & Kalverboer 1994). The patterns of the behaviour, such as social isolation, shyness and difficulties forming friendships, are similar to social behaviours described in children with an autistic spectrum disorder.

Children with a diagnosis of ADHD are also described as having difficulties in interaction with their peers although a primary diagnosis of autism is recommended by DSM 1V criteria, where these difficulties are significant (APA, 1994). Clark, Feehan, Tinline & Vostanis (1999) estimated the rates of autistic symptoms in a group of children with ADHD and concluded that a high percentage of parents (65-80%) reported significant difficulties with social interaction, particularly in empathy and peer relationships and communication, including imaginative ability, nonverbal communication and maintaining conversation. As mentioned, Gillberg (1989) has suggested links between motor problems

and ADHD and the continuum of autistic spectrum disorders. He suggested a continuum of disorders from the child with severe learning difficulties as described by Wing (1981b), Kanner's children with mild learning difficulties (Kanner 1943) and autism, Asperger syndrome and children with a diagnosis of DAMP (Gillberg & Gillberg 1989).

More recently, studies have begun to investigate the relationship between soft signs and psychiatric outcome and the relationship between DCD and Asperger syndrome (Pine, Wassermann, Fried, Parides & Shaffer, 1997; Kadesjo & Gillberg, 1999).

Rasmussen & Gillberg (2000) returned to the original groups reported in the series of studies, which investigated DCD and ADHD. They carried out a follow-up study of 55 subjects aged 22 years, who had ADHD with and without co-morbid DCD. The presence of ADHD with DCD, and DCD alone were important predictors of poor outlook. They found 69% of the severely affected ADHD and DCD group and 80% of the small group with DCD only, had poor outcomes. Eight children in the group with severe ADHD and DCD had an autism spectrum disorder when first examined at age 7 years. Four of these people had scores indicative of autism spectrum problems, measured by the ASD-I (Ehlers & Gillberg 1993) at follow-up, and two who had Asperger disorder at 7 years had ASD-I scores close to autism spectrum cut-off at follow-up.

1.4.0 Asperger Syndrome

The condition known, as Asperger syndrome was first described in 1944 (Asperger 1944; Frith 1991) and was brought to the attention of the English-speaking world by Wing (1981). Asperger syndrome is a neurodevelopmental disorder, which is defined by social deficits. Many of the clinical features closely resemble those seen in high functioning autism.

There has been considerable debate in the literature about whether the condition is on a continuum with autistic conditions and it is generally considered to be a form of autism in children with cognitive functioning at least within the average range. Since Wing's paper in 1981 there has been increasing interest in the diagnostic criteria, which provide the best description of Asperger syndrome. A number of authors (Gillberg & Gillberg, 1989; Szatmari, Bremner & Nagy, 1989; Wing, 1981) have suggested criteria and both ICD-10 and DSM-IV now include Asperger syndrome in their manual. As with autism the definitions are based on behavioural descriptions and one of the main areas of dispute has been over the presence or absence of significant language delay.

1.4.1 Diagnostic Criteria for Asperger Syndrome

The diagnostic guidelines listed by ICD -10 include:

- No clinically significant delay in spoken or receptive language or cognitive skills.
- Single words should have developed by age two and communicative phrases by age three
- Motor skills may be delayed and motor clumsiness is usual but is not a necessary diagnostic feature.
- Qualitative abnormalities in reciprocal social interaction
- Children have unusual and circumscribed interests or restricted repetitive or stereotyped patterns of behaviour interests and activities.
- The disorder cannot be attributed to other pervasive developmental disorders.

The DSM IV diagnostic criteria for Asperger's disorder include qualitative impairment in social interaction as manifested by at least two of the following:

- Marked impairment in the use of multiple non-verbal behaviours such as eye-to-eye gaze, or facial expression. Failure to develop peer relationships appropriate for

development level, and lack of spontaneous seeking to share enjoyment, interests or achievements with other people.

- Restricted repetitive and stereotyped patterns of behaviour, interests and activities, including encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal in intensity and focus. Stereotyped and repetitive motor mannerisms may also be present.
- Significant impairment in social, occupational and other areas of functioning
- No clinically significant delay in language or cognitive development.

Other criteria used include social impairment, pedantic speech, and obsessive interests (Gillberg & Gillberg, 1989; Szatmari, 1989). The child may make inappropriate approaches to their peers, and show intense interest in circumscribed topics, generally learnt in a rote manner.

Clumsy ill-co-ordinated movements, including difficulty with fine motor tasks, are usually a diagnostic feature of this group although neither the ICD-10 nor DSM-IV regards clumsiness to be a necessary diagnostic feature. Asperger mentioned motor clumsiness in the clinical cases he described although he did not include it in the overall clinical picture, which he presented. Wing, (1981) mentions difficulties with motor skills, reporting there is often a history of delayed acquisition of motor skills and difficulties with poor eye-hand co-ordination and visual-spatial skills (Wing 1981), but she does not include motor difficulties as a necessary feature for a diagnosis of Asperger syndrome.

Table 1.1 lists the range of criteria mentioned by different classification systems as associated with Asperger syndrome but not all are required for a diagnosis. The criteria where there is agreement include social impairment, and communication difficulties, and all absorbing interests. There is not agreement over the quality of language impairment and if

early language may be present. Although some authors include clumsiness or motor impairment as a diagnostic criterion, there have been no attempts in the literature to define the degree of motor impairment necessary to fulfil the requirement for difficulties with motor skills. Although ICD-10 and DSM-IV criteria are in agreement, these diagnostic criteria have not been used consistently in studies describing Asperger syndrome, and criteria need further validation.

1.1 Comparison of the Clinical Features of Asperger Syndrome

	Asperger 1944	Wing 1981	ICD-10	DSM-IV	Gillberg 1989	Szatmari 1989	Tantam 1988
Speech Delay	No	May be present	No	No	May be present	Not stated	Use language freely
Cognitive Delay	No	May be present	No	No	May be present	Not stated	May be present
Social Impairment, Poor reciprocal social interaction	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Clumsiness/motor impairment	Yes	May be present	May be present	May be present	Yes	Yes	Yes
Pedantic Speech	Yes	Yes	Not stated	Not stated	Yes	Yes	Yes
Restricted and repetitive interests	Yes	Yes	Yes	Yes	Yes	Yes	Yes

(Howlin 2000; Ghaziuddin, Tsai & Ghaziuddin 1992)

1.4.2 Prevalence rates of Asperger syndrome

A study of prevalence of autistic spectrum conditions in Cambridgeshire reported 57 per 10,000 (Scott, Baron-Cohen, Bolton & Brayne, 2002). The authors explain the change in previously reported rates, such as 4-5 per 10,000 for children with classic autism Fombonne (1999), may be the result of changing diagnostic criteria. Prevalence rates for Asperger syndrome from the Scott study are not clear, because they were not able to reclassify children with autistic spectrum disorder as Asperger syndrome where appropriate. More recently Keen & Ward (2004) analysed the prevalence of autistic spectrum disorder in autistic children in a single health district in England and found mean prevalence rates of 4 per 1000 births. They do not distinguish rates for Asperger syndrome. This study confirmed rates found by other studies such as that from the Centres for Disease

control for Brick Township, New Jersey (CDC, 2000), which reported a prevalence rate of 40 per 10,000 three to ten year olds with autistic disorder, and 67 per 10,000 when children with PPPD-NOS and Asperger syndrome were included. It is also consistent with the study by Charman and Baird (2002), which suggested an overall rate of 40 and 60 per 10,000 births.

Wing and Gould, (1979) carried out a study in which all children under the age of fifteen in one area of London were screened to identify early childhood psychosis. Wing (1981) reports two children in this study showed most of the characteristics of Asperger syndrome giving a rate of 0.6 per 10,000 and 1.1 per 10,000 who could have had a diagnosis of autism when young but came to resemble Asperger syndrome later. The survey did not include any children who were attending normal schools in the area or who had not come to that attention of the local medical and educational services in the area. They conclude that this prevalence rate is probably an underestimate (Wing, 1981). Wing (1981) suggests a prevalence rate of 15 boys to 4 girls, and 13 boys to 2 girls in a series of children she studied.

A population-based study of 6 year olds in Goteborg in 1977 by Gillberg, Rasmussen, Carlstrom, Svenson and Waldenstrom (1982), found 1.2% of the children who had cognitive functioning within the normal range, had a combination of severe motor impairment (not fulfilling criteria for cerebral palsy) and attention deficit disorder. Another 10% of the population showed motor impairment without attention deficit disorder. A psychiatrist examined the children at age seven years and eight children out of 21 children showed 'autistic type traits'. One child fulfilled DSM –111R criteria for autism. For their report on the epidemiology of Asperger syndrome (Gillberg & Gillberg, 1989), the remaining seven children were re-investigated and three boys were found to fulfil the

criteria for Asperger syndrome. Another four children fulfilled at least three of the criteria. This represents 26 in 10,000 of the population of six year olds. The children investigated in the study all had motor impairment.

Gillberg (1989) also reported a study by Holmström (1985) who screened all mainstream schools in Goteborg for children with autism or Asperger syndrome. A structured questionnaire for teachers and school staff, explored the presence of social impairment, language peculiarities, odd behaviour, autism type behaviour, and problems with academic achievement. Although the children and families were not interviewed the results indicated 8 in 10,000 children, aged 7-16, would receive a diagnosis of Asperger syndrome. This survey did not include children in special school or those who were attending specialist autistic units.

Ehlers and Gillberg (1993) carried out a population study for Asperger syndrome, and screened school children in an outer borough of Goteborg. They applied the Gillberg and Gillberg (1989) criteria, which included motor clumsiness. They used a screening questionnaire to identify children at risk and then carried out a comprehensive diagnostic assessment. Out of a typical school population of 1401 children they found a prevalence of 0.36% and a male female ratio of 4:1. When four suspected cases were included, the prevalence increased to 0.71%, and the male female ratio was 2:1. Ehlers & Gillberg also applied the Szatmari (1989) criteria and found a prevalence rate of 0.50% and when they applied ICD-10 criteria the prevalence for definite Asperger syndrome was 0.29%.

In 1999 Kadesjo, Gillberg, and Hagberg, (1999) screened 826 seven-year-old children living in Karlstad, Sweden. All children were screened for autistic spectrum disorders, which included autistic disorder, Asperger syndrome, and other autistic like conditions. They found a rate of 48 in 10,000 for Asperger syndrome.

More recently Webb, Thompsen, Morey, Fraser, Butler & Barber (2000) aimed to estimate the prevalence of Asperger syndrome and High Functioning Autism (HFA) in mainstream children. Out of 11,600 children screened 10 fulfilled the criteria for Asperger syndrome or HFA. The results gave a minimum prevalence of 1:1000 for AS/HFA in the population. Although they used the Autism Spectrum Screening Questionnaire (Ehlers & Gillberg 1993) they did not discriminate between Asperger syndrome and HFA and the results do not include children who may have been attending special schools.

1.4.3 Motor Impairment in Asperger Syndrome

The study by Gillberg of 23 children with Asperger syndrome (1989) found that all but 4 of 21 children tested with the Griffiths Developmental Scales were conspicuously 'clumsy' in motor skills where clumsiness was diagnosed as a score on the motor scale > 15 points below the child's IQ level. A further two children were judged clumsy on clinical grounds. That gives a rate of 84% of the children with Asperger syndrome with significant difficulties with motor skills. However it is not clear what diagnostic criteria were used for the initial diagnosis of Asperger syndrome, and the numbers in these groups are small.

Bonnet and Gao (1996) estimated 80% of subjects with Asperger syndrome display motor dyspraxia or clumsiness.

A study investigating the nature of motor impairment in Asperger syndrome, carried out by Green, et al., (2002) found that motor impairment was universal in their sample of 11 boys with Asperger syndrome, with the scores of all boys falling below the 15th percentile on the Movement ABC (Henderson & Sugden 1992).

Burgoine & Wing (1983), Tantam (1988) and Gillberg (1989) have all proposed clumsiness as an essential diagnostic feature of Asperger syndrome. As with the definition of motor impairment in DCD, the definition of clumsiness has not been clarified, and there has not

been consensus on whether these difficulties are referring to fine motor or gross motor skills, or whether DCD provides an adequate definition of the motor difficulties observed in children with Asperger syndrome. The cases described by Asperger (1944) were said to be poor at games involving motor skills and were reported to have difficulty with writing and drawing.

A review of the literature on clumsiness as a diagnostic feature of Asperger Syndrome by Ghaziuddin, Tsai & Ghaziuddin, (1992) identified 42 papers with the term Asperger Syndrome in the title. They categorised papers into those, which defined clumsiness and dealt specifically with the symptom, those that listed clumsiness as a feature of Asperger syndrome, and those who used standardised tests to describe clumsiness. Ghaziuddin et al (1992) found that none of these papers had defined clumsiness. Few papers had used clumsiness as a diagnostic criterion and others mentioned it as an incidental finding. Only four papers had used tests to define clumsiness, no study offered an operational definition of clumsiness, and the relationship to IQ was not considered. They found that studies did not differentiate between autism and Asperger syndrome and it appeared that in the studies that did attempt to measure clumsiness the rationale for the specific test used was not explained. Ghaziuddin et al (1992) concluded that in many cases the decision on the presence of clumsiness was based on the clinician's impression. They did not differentiate between different types of clumsiness, for example fine motor or gross motor impairment, and did not consider the effect of age. Ghaziuddin and colleagues (1992) concluded the symptom had not been adequately studied. They recommended that standardised tests should be used to provide an operational definition of clumsiness and that age and IQ should be taken into account.

Following this review Ghaziuddin, Butler, Tsai, and Ghaziuddin, (1994) carried out a study to evaluate the presence of clumsiness in young children and adolescents with Asperger syndrome, and compared it to that occurring in young people with High Functioning Autism (HFA). The Bruininks-Oseretsky test (Bruininks 1978) was administered to 10 children with Asperger syndrome and 9 with HFA to assess gross and fine motor skills. Deficits in motor functioning were present in both groups when compared with age matched population norms, and they concluded that clumsiness is not unique to Asperger syndrome, and that the presence of low IQ must be taken into account. They also suggested that the clumsiness may not only be due to poor motor coordination, but also information processing deficits, or difficulties in visual perception (Ghaziuddin, Butler, Tsai, & Ghaziuddin, 1994).

Ghaziuddin and Butler (1998) then investigated the presence of clumsiness in patients with autism, Asperger syndrome and PDDNOS, who had all been assessed on a standardised diagnostic protocol and diagnosed on the basis of DSM-IV and ICD-10 criteria. All patients with pervasive developmental disorders in the study showed problems with motor coordination. Those with a diagnosis of autism were found to be most clumsy, followed by PPDNOS and Asperger syndrome. Ghaziuddin and Butler (1998) recommended the need for caution before accepting clumsiness as a specific diagnostic criterion for Asperger syndrome. They also recommended that attempts should be made to separate cognitive from motor aspects but did not rule out the possibility that the pattern of motor deficits in Asperger syndrome might be different from that seen in autism.

A study, which examined the external validity of HFA and Asperger syndrome carried out by Miller and Ozonoff, (2000) compared the neuropsychological domains thought to discriminate between the two groups. These included intellectual functioning, early

language history, motor ability, visuo-spatial skills and executive functions. When group differences in IQ were controlled for, a marginally significant trend toward greater impairment in motor skills was found in the Asperger syndrome group. However, they also found that both groups showed below average motor skills when compared to population norms.

From this series of investigations it would seem that clumsiness is a feature of autistic spectrum disorders, but is not specific to Asperger syndrome. Recent studies, (Weimer, Schatz, Lincoln, Ballantyne & Trauner 2001; Green et al., 2002) have begun to look more specifically at the nature of motor impairments in children with Asperger syndrome. For example, Hughes (1996) looked at planning problems in children with autism, and reported problems in executing goal-directed motor acts, which are apparent in even very simple situations. No distinction was made between children with autism or Asperger syndrome.

To further investigate the nature of the motor dysfunction, Weimer Schatz, Lincoln, Ballantyne & Trauner (2001) compared subjects with Asperger syndrome with those with no neurological impairment. Subjects were matched on age, sex, socio-economic status and Verbal IQ. A range of motor tests was administered to assess motor speed, fine motor control, motor planning, balance, visuo-motor function and praxis (the ability to carry out skilled, purposeful and coordinated motor activity). The mean age of the group was 15.7 years. Asperger syndrome was diagnosed using DSM-IV criteria. Those with Asperger syndrome had significantly worse performance on tests of balance, and on serial finger-thumb apposition. The Apraxia test indicated that Asperger syndrome groups performed significantly more poorly on whole body postures and on total scores, than the normal subjects. Weimer et al (2001) concluded that the motor impairment or clumsiness observed in Asperger syndrome may be sensory deficits rather than motor deficits and suggest that

such deficits can explain the clumsy appearance of people with Asperger syndrome and would explain difficulties with catching, throwing and kicking which are often described.

The motor performance of an older group of autistic people was compared with that of people with learning difficulties (Morin & Reid 1985) matched on age and IQ. They report a trend for inferior qualitative scores by autistic people, but there was no quantitative difference between the groups, which lead them to conclude that poor motor performance is a factor of mental retardation. The motor patterns in throwing, jumping and running of the autistic individuals were described as immature with inappropriate and non-functional arm movements being common.

Green et al (2002) used the Movement ABC (Henderson & Sugden 1992) to quantify the extent and severity of motor difficulties among the children with Asperger syndrome, and to explore whether the motor difficulties were distinctive and 'syndrome specific'. They found that the Asperger syndrome group had higher mean impairment scores than a group with Specific Developmental Disorder of Motor function (SDD-MF) and that the overall trend towards greater impairment in the Asperger syndrome group approached significance. Tasks involving aiming and catching a ball best distinguished the Asperger syndrome boys from the SDD-MF boys. They concluded that motor impairment might be almost universal in Asperger syndrome, although the severity of impairment is variable.

There is therefore agreement in the literature that motor impairment is a feature commonly seen in Asperger syndrome, but although there have been studies investigating the specific nature of these difficulties suggesting problems with jumping throwing and aiming and catching a ball, as yet there is no agreement that these difficulties are diagnostic of Asperger syndrome as opposed to other autistic disorders.

1.4.4 Behavioural and emotional disturbance in Asperger Syndrome

There has also been a body of literature commenting on and investigating the occurrence of psychiatric disorders in people with a diagnosis of Asperger syndrome. Neumann and Walker (1996) reviewed the literature on childhood soft motor signs, behavioural difficulties and adult psychiatric difficulty including depression and schizophrenia. They investigated precursors of mental illness and following examination of childhood home movies found a high rate of motor skills deficits and neuromotor abnormalities in young children who later develop adult-onset schizophrenia. They made a differentiation between neuromotor abnormalities and motor skills impairment.

The literature has begun to investigate the differences in long-term outcome in children with Asperger syndrome and High Functioning Autism with particular reference to emotional and adaptive behaviour.

To investigate behavioural and emotional disturbance in high functioning autism and Asperger syndrome, Tonge, Brereton Gray and Einfeld (1999) assessed psychopathology in 75 children and adolescents with autism (mean age 7.41 years) and 52 with Asperger syndrome (DSM-IV criteria) (mean age 9.89 years), using the Developmental Behaviour Checklist (Einfeld & Tonge 1994). The checklist is designed to assess a range of behavioural and emotional problems of children with intellectual disability. They found that 65 percent of the HFA group and 85 percent of Asperger syndrome group showed clinically significant levels of behavioural and emotional disturbance. The Asperger syndrome group scored significantly higher than the HFA group on overall severity of psychopathology, and on the disruptive behaviour, anxiety, autistic/social relating and antisocial scales.

A review of the literature by Kim, Szatmari, Bryson, Streiner and Wilson (2000) compared the prevalence and characteristics of anxiety and mood problems among Asperger syndrome children and autistic children. They found several reports that measured symptoms of anxiety and mood among PDD (pervasive developmental disorders) children and found higher than expected scores than among other groups. The studies have not attempted to differentiate anxiety / mood symptoms from PDD symptoms. Kim et al (2000) investigated the prevalence of co-morbid psychiatric symptoms in children with PDD and aimed to assess the impact of these additional difficulties on overall outcome. They investigated 47 children who met criteria for autism, (DSM-IV criteria) and 21 who met criteria for Asperger syndrome (DSM-IV criteria). Children were enrolled in the study at age 4 to 5 years (mean age 5 years 6 months) when they were given a full psychometric assessment and a semi-structured interview was carried out with the parents. They were then followed up six years later when they were between 9 and 14 years (mean age 12 years). At follow-up a questionnaire (OCHS-R 1983, Boyle, et al., 1993) was administered to parents to measure the prevalence of psychiatric problems. When compared with a random sample of community children, 16.9 percent of the PDD children scored at least two standard deviations above the population mean on a measure of ADHD, 16.9 percent scored at the same level on a measure of depression and 13.6 percent scored at the same level on a measure of generalised anxiety. On a measure of internalising behaviour (over-anxious, depression and separation anxiety) 13.6 percent were classified 'clinically relevant'. Children with anxiety and mood problems were also found to have more aggressive and oppositional behaviour. Mood problems seemed to have a significant impact on family life and social relationships (Kim et. al. 2000). Kim and colleagues also reported that measures of 'depression' and 'generalised anxiety' did not differ significantly between

children diagnosed with Asperger syndrome or autism. However they concluded that co-morbid psychiatric problems are more common in children with PDD than in a community population. Clinically relevant depression was the most common problem and these problems were found to have a significant effect on the lives of the children and their parents (Kim et.al.2000).

An investigation looking at co-morbidity of Asperger syndrome by Ghaziuddin and colleagues (Ghaziuddin, Weidmer-Mikhail & Ghaziuddin, 1998) found that in children aged between 6 to 12 years 50 percent had a diagnosis of ADHD, or depression, and one child had both. In the group over 13 years, which included adults, 11 out of 15 people had a diagnosis of depression, ADHD, obsessive-compulsive disorder, Tourette's syndrome or tic disorder.

There are therefore a range of diagnoses which have been found to co-occur with Asperger syndrome and which have long term implications for the child's future functioning. However, where studies have aimed to compare autism and Asperger syndrome, emotional and behavioural outcomes did not differentiate between the two diagnostic categories although there is a suggestion that severity of emotional and behavioural disorder may discriminate between the two groups (Tonge et al. 1999).

1.5 Conclusions

Investigations into Developmental Coordination Disorder and Asperger syndrome, suggest areas of overlap, both in diagnostic criteria and in descriptive features. Asperger syndrome has been strongly associated with motor impairment, and the literature reports that up to 80% of children with Asperger syndrome display motor difficulties (Gillberg 1989; Bonnet & Gao 1996). The literature has also begun to investigate more specific aspects of motor impairment seen in children with Asperger syndrome, but there has been no attempt to

quantify the degree of motor impairment required to fulfil diagnostic criterion (Green et al 2002). However, there is continuing debate in the literature about the basic criteria for a diagnosis of Asperger syndrome, and there is disagreement over whether motor impairment is a necessary diagnostic criterion. Studies have indicated that motor impairment, a feature commonly seen in children with an autistic spectrum diagnosis, does not specifically differentiate children with Asperger syndrome (Ghaziuddin, Butler, Tsai, & Ghaziuddin 1994; Ghaziuddin & Butler 1998; Miller & Ozonoff 2000).

There is also increasing evidence of social and emotional disturbance and other behavioural difficulties in children with DCD and children with Asperger syndrome, indicating there may be links between problems with motor impairment and behavioural and emotional difficulties. Gillberg suggests that there are a wide range of behaviour and emotional difficulties associated with motor problems in children including autistic like behaviour. Difficulties with social interaction are a diagnostic criterion of autistic spectrum disorders and Schoemaker & Kalverboer (1994) for example concluded that all children in their study of children with DCD had social and affective problems irrespective of degree of motor impairment. However there has been no attempt to define the nature of behavioural difficulties seen in DCD or to describe them as an essential feature of the diagnostic criteria.

Studies such as that by Kadesjo & Gillberg (1999) have found children with DCD had more symptoms of Asperger syndrome than children without DCD and found that there was a strong interactive effect of DCD and ADHD and strong correlation with Asperger syndrome. Other researchers such as Green et al (2002) have begun to report on the incidence of DCD and patterns of motor impairment in children with Asperger syndrome, but have not looked at the presence of autistic behaviour in a group of children with DCD.

Studies (Kim et al 2000; Ghaziuddin et al 1998) investigating the presence of behaviour and emotional difficulties in children with Asperger syndrome found problems with depression, ADHD, and anxiety were much more frequent than in a typical population, although as with motor impairment these do not discriminate between Asperger syndrome and other autistic spectrum disorders. There are also results from long-term studies of children with motor signs that these associated emotional and behavioural difficulties are not short term. However, motor difficulties are reported in children with a range of developmental conditions, for example children with low birth weight (Marlow et al 1993) and as yet it is not clear if difficulties with behaviour and social impairment are consequent on the motor impairment or are part of a wider picture of impairment generally seen in children with developmental difficulties.

In summary, there are patterns of difficulties with behaviour and emotion that are common to children with DCD and children with autistic spectrum disorders and the long term outcome for both groups indicates a high risk of co-morbid psychopathology, particularly depression and anxiety, and ADHD.

The studies reviewed here have looked at the relationship between motor impairment and specific aspects of emotional and behavioural development, such as ADHD. However, few studies investigated the presence and range of emotional and behavioural difficulties shown by children with DCD nor have studies investigated the association between specific aspects of behaviour, and the degree of motor impairment, developmental level and age in the same group of children. Gillberg identified children with Asperger syndrome in his cohort of children with DCD and attention deficit disorder and Hellgren et al (1994) found a high incidence of social negativism and social withdrawal in the same cohort when they were reassessed at age sixteen. The children with a diagnosis of Asperger syndrome in the

group had also had attention deficit disorder diagnosed at age seven and were not identified from the total cohort of children with DCD. The series of studies did not aim to screen all the children with DCD for a diagnosis of Asperger syndrome but did identify a small group of children with autistic traits.

Despite the indications in the literature that children with DCD are likely to show difficulties with social interaction and that motor impairment is characteristic of children with autistic spectrum disorder, there have been few studies, which have aimed to assess specifically the presence of autistic behaviour in a group of children with DCD.

The present study aims to fill this gap by investigating in more detail the nature of the association and will look at the range of behaviour and emotional problems that children with DCD may exhibit, with particular reference to autistic spectrum disorder.

1.6 Hypotheses

Analysis of the data was structured to address a number of hypotheses which together aim to clarify the relationship between motor skills, behavioural difficulties and autistic like behaviour.

In the first instance children were grouped according to degree of motor impairment and comparisons among group means were made.

Hypotheses investigated were as follows:

1. When allocated to groups according to the degree of motor impairment, children with severe DCD and moderate DCD would have higher mean scores on a standardised measure of behaviour than children without motor impairment.
2. Those children with motor impairment would have high hyperactivity scores and high unsociability scores on a measure of behaviour.

3. Those children with children with high levels of clinical behaviour problems as defined by a standardised measure of behaviour would have high scores on a measure of motor impairment.
4. Those children with motor impairment would be more likely to have high scores on a measure of autistic like behaviour
5. Those children with high scores on a measure of autistic like behaviour would have high scores on a measure of motor impairment.
6. There would be differences across the groups allocated on basis of DCD and presence or otherwise of clinical behaviour problems, on the measure of autistic like behaviour.

A subsidiary research question investigated the pattern of reported motor difficulty related to degree of behaviour problems or to autistic like behaviour. The hypothesis investigated whether children who had clinical behaviour problems show specific areas of motor difficulty. This subsidiary question also investigated the hypothesis that children with high levels of autistic like behaviour would show specific areas of motor difficulty?

To further test out the hypothesis that there would be a relationship between motor impairment, clinical behaviour problems, and autistic like behaviour, data were further analysed by the use of correlations to investigate relationships among measures. The following hypotheses were examined.

1. There would be a relationship between the degree of motor impairment and difficulties with behaviour measured.
2. There would be a relationship between the degree of motor impairment and autistic like behaviour.

3. There would be correlations among measures of motor impairment, behaviour, and autistic like behaviour. Specifically children with motor impairment would be more likely to have high scores on measures of behaviour and autistic like behaviour.

As a subsidiary research question, partial correlations were carried out to investigate the effects of age and IQ on the relationship among the three variables of motor skills, behaviour and autistic like behaviour.

CHAPTER 2

EMPIRICAL STUDY

2.1 METHOD

2.1.1 Participants

The sample used in this study was recruited from the Neurodevelopmental Assessment Clinic of the Neurodisability Service, Great Ormond Street Hospital, the Dyspraxia Foundation and through personal, informal contact. The children were aged between 6 years and 12 years. The study had ethical approval from The Great Ormond Street Hospital for Children NHS Trust/Institute of Child Health Local Research Ethics Committee (Appendix 1). One hundred and eleven families were approached to take part in the study and one hundred and seven families gave their informed consent after full explanation of the study.

Of these, 54 children were recruited from the Neurodevelopmental Assessment Clinic. Referral letters were reviewed and where there was a question related to motor function, selected families were approached directly when they attended for first appointment. Following the initial family interview with the multidisciplinary team, the parents were approached personally by the researcher and recruited to the study. Although an attempt was made to enlist all children who fulfilled the recruitment criteria, recruitment was incomplete. Families were not approached if the multi-disciplinary team identified significant emotional or psychological difficulties indicating an approach would be inappropriate and could significantly influence the process of the clinical work. Families may also have been missed if motor difficulties were not described either in the referral letter or in the initial interview. It is possible other families were not approached because for practical reasons the researcher was not available to monitor every child attending the

clinic. All families who were approached through the Neurodevelopmental Assessment Clinic agreed to take part.

Another group of children was recruited through the Dyspraxia Foundation, following an advertisement in that society's newsletter and on its web site asking for volunteers. Thirty-three families responded to the advertisement in the Dyspraxia Foundation newsletter or the web site, and 29 families completed the questionnaires. Parents were asked to write or phone the researcher. These children had been diagnosed with developmental coordination disorder by local services.

A further group of 24 children with no motor difficulties who were attending mainstream school were recruited both from the neurodevelopmental assessment clinic and through personal contact. All families of children with no motor difficulties who were approached agreed to take part in the study.

Overall the sample is a mixture of a clinic and opportunity sample.

Several criteria were used to select children from the clinic population. Children with obvious organic pathology, congenital disorder, e.g. cerebral palsy, a history of brain injury, or known genetic diagnosis were excluded. These conditions were routinely mentioned in referral letters as part of the referral question. Children were also excluded if the referral question included the possibility of a genetic diagnosis such as Fragile X. The clinic sample had all had genetic testing and any children with positive results were excluded. It is not known how many of the non-clinic sample had had genetic testing or other pathology, as this information was not available.

No children in the study were known to have a diagnosis of autistic spectrum disorder (ASD) when recruited

2.1.2 Procedure

All families who agreed to take part in the study were given an information sheet explaining the aims and procedure of the study and were asked to sign a consent form. The consent form included information on complaints procedure and contact information for the Chairman of the Research Ethics Committee, Great Ormond Street Hospital and Institute of Child Health. (Appendix 1)

Data were gathered by use of parent report and psychological assessment. Parents who attended the Neurodevelopmental Assessment Clinic were asked to complete a series of questionnaires either during waiting time at the clinic or following the appointment. All families who were approached in this way completed the questionnaires, and also agreed to psychological assessment. The researcher was available for questions if necessary, and to ensure questionnaires were completed. Families who responded through the Dyspraxia Foundation were sent questionnaires by post with the facility to contact the researcher by telephone or mail with questions if necessary. A self-addressed envelope was provided for return of the questionnaires. Several parents contacted the researcher by phone but no parents made contact to discuss problems completing the questionnaires.

2.2 Materials

2.2.1 Movement Assessment Battery Checklist

The Movement Assessment Battery Checklist (Sugden and Henderson 1992) was developed to provide an observational style of assessment, and to complement the formal procedure of the Movement Assessment Battery test. The checklist was designed to be completed by an adult familiar with the child's day-to-day motor functioning and to assess the child's functioning in everyday situations. It focuses on the qualitative aspects of the child's motor performance and on emotional and behavioural aspects. The checklist

consists of 48 items divided into four sections, each of which considers the child's motor performance in progressively more complex situations. The total possible score is 144. The items reflect the motor activities common in the day-to-day life of 4-12 year olds. An additional section for behaviour problems was designed to consider the extent to which a child's attitudes and feelings about motor tasks are situation specific or more generalised. The behaviour section related to motor difficulties is not used in the calculation of scores to identify motor difficulties. The sections are as follows:

Section 1: Child Stationary/Environment Stable,

Section 2: Child Moving/Environment Stable,

Section 3: Child Stationary/ Environment Changing

Section 4: Child Moving/Environment Changing

Section 5: Behavioural Problems Related to Motor Difficulties.

The checklist can be used to identify a child with motor difficulties by comparing the child's score to the standardized norms for other children in the same age group. Total scores on the motor component of the Checklist can be transformed to percentiles and used to give cut-off points for children in the age ranges from 6 years, 7 years, 8 years and 9 + years, the higher the percentiles the better the performance. Results can be divided into a normal performing group, scores $> 15^{\text{th}}$ percentile, for a borderline group with scores between the 6^{th} and 15^{th} percentile and for the 5% of the population with the most severe movement problems i.e. scores $\leq 5^{\text{th}}$ percentile. Profiles of motor skills, provided by the Movement Assessment Battery Checklist could also be used to look specifically at patterns of motor difficulty.

Wright and Sugden (1996) carried out a two-step procedure for the identification of children with developmental coordination disorder in children in Singapore and found that

of 64 children who were selected by the ABC Checklist as having movement difficulties, either with scores below the fifteenth percentile or below the fifth percentile, seventeen were confirmed as to category by the ABC test, a further three children with borderline scores were confirmed to have movement difficulties by the ABC test and 22 of the remaining 47 children who were identified by the checklist had high scores on the ABC test indicating motor difficulty. Wright and Sugden (1996) also provide UK data on levels of agreement between the two measures, reporting that eleven out of sixteen children selected by the 5th percentile cut-off on the checklist were confirmed as having movement difficulties. Another study (Schoemaker, Smits-Engelsman & Jongmans 2003) has also looked at standards of reliability and validity for the ABC-Checklist and recommended its use by teachers as a screening tool for children with movement difficulties. No studies have reported its use with parents. Several children in the present study were also assessed on the Movement ABC Test, and the results in each case confirmed the diagnosis of Developmental Coordination Disorder. These results have not been analysed in detail for the present study, as only children who were part of the clinic sample had further assessment results available and a full set of data was not available. However it can be reasonably assumed that children with high scores on the ABC Checklist do have motor difficulties and that the cut-off points represent suitable criteria for a diagnosis of Developmental Coordination Disorder based on ICD-10 criteria, and DSM-IV criteria. When recruited to the study no children had a primary diagnosis of pervasive developmental disorder.

2.2.1.1 Group Allocation

Children were allocated to one of three groups according their results on the ABC Checklist, identified by the 5th and 15th percentile cut-off criteria.

GROUP 1: Developmental Coordination Disorder Severe

This group consisted of children identified by the 5th percentile cut-off, indicating severe problems with motor coordination.

GROUP 2: Developmental Coordination Disorder Moderate

This group consisted of those children identified by the 15th percentile cut-off, indicating moderate motor coordination difficulties.

GROUP 3: No Developmental Coordination Disorder

This group consisted of those children whose results were not identified by the 15th percentile cut-off indicating no difficulties with motor coordination.

2.2.2 Rutter A (2) Children's Behaviour Questionnaire

The Rutter A (2) scale (Rutter, Tizard & Whitmore, 1970; Schachar, Rutter, & Smith, 1981; Elander and Rutter 1996) is a children's behaviour questionnaire developed to discriminate between different types of behavioural and emotional disorders and to discriminate between children who show disorders and those who do not. It was developed for completion by parents. The questionnaire is long established and has proved to be valid and reliable in a variety of contexts (Elander & Rutter 1996; Hogg, Rutter & Richman 1997). It consists of a number behavioural descriptions for each of which the informant is asked to mark 'doesn't apply' 'applies somewhat' or 'certainly applies'. Replies are given a weight of 0, 1, and 2 respectively and scores of individual items are added to give a final score. The total possible score is 62, and a cut-off point of 13 or more has been identified (Rutter, Tizard, and Whitmore, 1970) which discriminates between children with and without clinical behaviour problems.

Scores from selected items can be used to discriminate between emotional, conduct and hyperactivity disorder. Schachar, Rutter & Smith (1981) also identified an unsociability

index. As hyperactivity (Gillberg 1989) and unsociability have been specifically mentioned in the literature in connection with both DCD and Asperger syndrome, these two indices were investigated for the present study. Conduct disorder and emotional disorder have also been mentioned in the literature on children with DCD, but were not investigated, although the information was available, because the focus of the study was on social interaction and hyperactivity. Items contributing to the hyperactivity index include 1) 'very restless, often running about or jumping up and down, hardly ever still' 2) 'Squirmy fidgety child' and 3) 'cannot settle to anything for more than a few moments'. A score with a range from 0 – 6 can be obtained for this factor by adding the scores of the three individual items. Hyperactivity is considered present when the score is 3 or more.

Poor peer relationships can be examined to provide an unsociability score. Items contributing to this factor include 1) 'frequently fights with other children' 2) 'not much liked by other children' and 3) 'Tends to do things on his own, rather solitary'. The scores range from 0 – 6. Unsociability is considered present when the score is 3 or more.

2.2.3 Adapted Autism Spectrum Screening Questionnaire (Gillberg 1993, 1999)

An adapted version of the Autism Spectrum Screening Questionnaire (ASSQ) was used to assess the presence of autistic like behaviour. Ehlers and Gillberg (1993) first described the questionnaire in a study investigating the incidence of Asperger Syndrome in a total population in an outer Goteborgborough. The questionnaire was specifically designed for teacher screening as part of the population study of Asperger syndrome in a mixed group of 1519 seven to sixteen year-old children living in the area in March 1991. A full questionnaire was completed for a total of 1401 children. The items included were considered to reflect impairments in social interaction, communication behaviour and

circumscribed interests, as characteristic of Asperger syndrome, and based on Gillberg and Gillberg (1991) criteria. The original scale consists of a 27-item checklist for completion by lay informants, for use with children and adolescents with normal intelligence or mild learning difficulties. Further work on reliability and validity for a teacher and parent version has been carried out by Ehlers Gillberg & Wing (1999). They reported that a cut off score of 19 for parent rating correctly identified 82% of the children functioning within the autistic spectrum, although the scale did not differentiate between those with high functioning autism and Asperger syndrome. Following an evaluation of the pattern of responses across the main categories according to Gillberg's criteria, 16 items were considered most characteristic of Asperger syndrome (Ehlers & Gillberg 1993).

The questionnaire was also used in a total population study by Kadesjo, Gillberg & Hagberg (1999) when a score of 17 or more identified children with Asperger syndrome. Only one other study by Webb, Thompsen, Morey, Fraser, Butler & Barber (2003) has reported using the ASSQ as part of a study to estimate the prevalence of Asperger syndrome and high-functioning autism in mainstream children. They found an ASSQ score greater than 21 was predictive for Autistic spectrum.

For the purposes of the present study, a shorter version of the questionnaire was used. This version was first developed by H. McConachie (1996) (personal communication) as part of the process of developing assessment procedures for children attending the neurodevelopmental assessment clinic at the Wolfson Centre, Great Ormond Street Hospital for Children, London. The brief version of the scale includes 15 items, which according to Gillberg's criteria were considered most characteristic of Asperger Syndrome (Ehlers and Gillberg 1993). Because the study aims to investigate links between motor difficulties and autistic spectrum disorders, the question 'has clumsy, ill coordinated

ungainly awkward movements or gestures' was not included in the analysis despite being one of the items considered by Gillberg et al (1993) most specific to Asperger Syndrome. Following concerns expressed by the Ethics Committee at Great Ormond Street Hospital about the language used in the original questionnaire, some changes were made to the questions to make them more easily understood by parents. The rater is asked to mark each item to describe how 'this child stands out as different from other children of his/her age in the following way:' Replies are given a weight of 0 (No), 1 (somewhat), and 2 (yes) respectively and scores of individual items are added to give a final score. The brief version has a total possible score of 30. The adapted questionnaire is called the AASSQ. However the changes to the original questionnaire mean that there is no information on validity and reliability. Although the original questionnaire has been shown not to distinguish between High Functioning Autism, and Asperger syndrome, the questions used are those Gillberg and Ehlers (1993) found to be most characteristic of Asperger syndrome, and it was expected would provide some evidence of the presence of autistic like behaviours.

The mean AASSQ score for the present sample is 11.55 (SD 8.23) and the median score is 11. As there is no recommended cut-off score for this adapted version of the ASSQ, and although not all children who score above the mean or median will fulfil the criteria for an autistic spectrum disorder, the median score of 11 as a measure of central tendency has been used as a cut off score for the purposes of this study. Because a score of eleven represents both the mean and the mid point of the distribution, it was concluded that scores above this point would indicate the presence of behavioural difficulties characteristic of autistic spectrum disorders.

2.2.4 Wechsler Intelligence Scale – Third Edition^{UK}

The Wechsler Intelligence Scale for Children - Third Edition^{UK} (Wechsler 1992) was administered to a sub-group of children. The scale was administered in full, following standard procedures, to allow calculation of Verbal IQ (VIQ) Performance IQ (PIQ), Full Scale IQ (FSIQ) and Index scores. The Wechsler Scales were used to provide a profile of cognitive functioning.

A full WISC-111 assessment was completed on 38 children, including 15 in Group 1 (Severe DCD), 5 in Group 2 (Moderate DCD) and 18 in Group 3 (no DCD). Time constraints and distance prevented complete collection of this data.

The ABC Checklist, Rutter A (2) Scales, and the AASSQ were completed by parents of 107 children.

CHAPTER 3

RESULTS

3.1 Characteristics of the Groups

The children were allocated to three groups based on degree of motor difficulty as measured by the ABC Checklist. The composition of the groups based on selection criteria is shown in Table 3.1.

TABLE 3.1 Characteristics of Groups

GROUP	Age Mean (SD)	Gender Male/Female	ABC Checklist Maximum Score: 144 Mean (SD)	Referral Source Dyspraxia, other /Neurodisability
Group 1 DCD-Severe N = 44	8.6 (1.5)	30/14	80.57 (18.36)	17/27
Group 2 DCD-Moderate N = 19	8.6 (1.9)	15/4	56.11 (18.62)	10/9
Group 3 No DCD N = 44	9.3 (2.1)	36/8	16.91 (2.27)	26/18
TOTAL N = 107	8.9 (1.8)	81/26	50.05 (33.71)	53/54

3.1.1 Age

The mean and standard deviations for the chronological ages of the three groups are set out in Table 3.1. Analysis of the means using one-way ANOVA shows homogeneous variances (Levene's $F = 3.39$, $df = 2, 104$, ns) and no significant difference between groups in Chronological Age ($F = 1.69$, $df = 2, 104$, ns).

3.1.2 Gender

The gender distribution of the three groups is shown in Table 3.1. Analysis of the data revealed no significant difference in male/female ratio across groups ($\chi^2 = 2.35$, $df = 2$, ns). Inspection of Table 3.1 shows there are more males than females in all three groups.

Although there are differences between the number of males and females in each group, the pattern is similar in the three groups and is consistent with that shown in epidemiological studies of the prevalence rates of behaviour disorder, Developmental Coordination Disorder in males and females, and in prevalence rates for males and females attending developmental assessment clinics (Gillberg 1995). The referral rates to the Developmental Assessment Clinic in the Neurodisability Service where part of this sample was recruited were 175 males and 50 females in the year 2000-2001 (Clinical Data).

3.1.3 ABC Checklist

To investigate the extent of the differences between the three groups on the ABC checklist, a one-way ANOVA was carried out on the data in Table 3.2.

TABLE 3.2: ABC Checklist Total Score and Section Scores by Group (DCD)

	ABC Checklist Maximum Score: 144 Mean (SD)	ABC Checklist Section 1 Mean (SD)	ABC Checklist Section 2 Mean (SD)	ABC Checklist Section 3 Mean (SD)	ABC Checklist Section 4 Mean (SD)
Group 1 DCD Severe N = 44	80.57 (18.36)	16.95 (6.37)	19.09 (5.57)	21.51 (6.04)	22.84 (5.49)
Group 2 DCD Moderate N = 19	56.11 (18.62)	14.22 (4.49)	12.94 (6.24)	14.39 (5.97)	15.94 (6.87)
Group 3 No DCD N = 44	16.91 (2.27)	4.93 (5.30)	2.98 (3.92)	4.47 (4.47)	4.77 (5.30)

Section 1 Child Stationary/Environment Stationary
Section 2 Child Moving /Environment Stable

Section 3 Child Stationary/Environment Changing
Section 4 Child Moving/Environment Changing

This revealed homogeneous variances (Levene's $F = 0.95$, d.f. = 2,104, ns). Because children were assigned to groups on the basis of scores on the checklist, a difference between groups was anticipated, and as expected one-way ANOVA indicated significant differences in means ($F = 153.69$, d.f. = 2,104 $p < .001$). Inspection of the data indicates the two groups of children above the percentile cut-offs have higher scores than the other group in line with the method of group allocation. Post hoc Scheffe tests showed a

significant difference in means between Groups 1 (Severe DCD) and Group 2 (Moderate DCD), Group 1 (Severe DCD) and 3 (No DCD), and Groups 2 (Moderate DCD) and 3 (No DCD), i.e. between groups with severe and moderate DCD, and between both groups with DCD and the group without DCD when compared separately.

To investigate the extent of differences among the four sections of the ABC checklist a one-way ANOVA was carried out on the data from the separate sections in Table 3.3. This revealed homogeneous variances (Levene's $F = 1.47$, d.f. = 2,104, ns; $F = 1.83$, d.f. = 2,101 ns; $F = 1.67$ d.f. = 2,104 ns; $F = 1.99$ d.f. = 2,101 ns) for sections 1, 2, 3, and 4 respectively. As expected one-way ANOVA indicated significant differences between group means ($F(2,101) = 51.1$ $p < .001$; $F(2,101) = 109.03$ $p < .001$; $F(2,101) = 106.31$ $p < .001$; $F(2,101) = 110.26$ $p < .001$). Inspection of the data indicates the two groups of children selected by the 5th and 15th percentile cut-offs have higher scores on each section than the children without DCD, in line with the method of group allocation. Post hoc Scheffe tests showed a significant difference in means between Groups 1 (Severe DCD) and Group 2 (Moderate DCD), on Sections 2, 3 and 4, but not on Section 1.

On each section, post hoc Scheffe tests also showed a significant difference in means between Group 1 (Severe DCD) and Group 3 (No DCD), and Group 2 (Moderate DCD), and Group 3 (No DCD), i.e. between both groups with DCD and the group without DCD when compared separately. Section 1, which describes the situation where the child is stationary and the environment is stable, is the only section where there was not a significant difference in mean scores between Groups 1 (Severe DCD) and 2 (Moderate DCD). This section includes items such as dressing, tying shoelaces, doing up buttons, personal hygiene, such as face and hand washing, differentiating left and right and recognizing body parts. The highest mean scores for Groups 1 (Severe DCD) and Group 2

(Moderate DCD) are in Section 4, which describes the situation where the child is moving and the environment is changing. This section includes items such as moving around the classroom, running to catch or kick a ball and other ball skills.

3.2 Comparisons of group means and frequencies

3.2.1 Rutter A (2) Scales

To investigate the first hypothesis that children with more severe motor impairment would have higher mean scores on measures of behaviour, specifically the Rutter A (2) a series of analyses were carried out.

The means and standard deviations for measures providing a description of behaviour as recorded on the Rutter A (2) scales across the three groups are shown in Table 3.3.

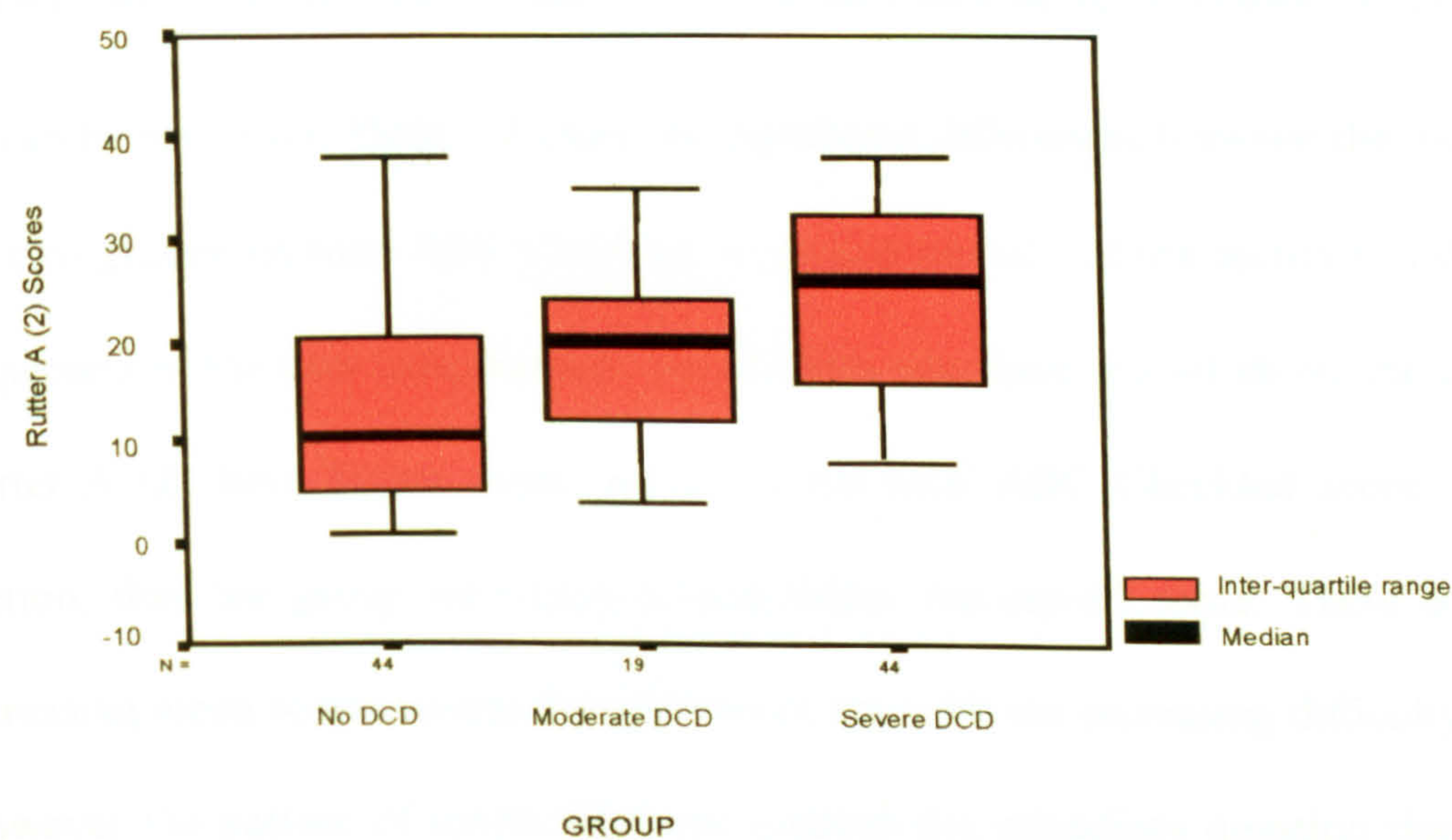
TABLE 3.3: Rutter A (2) scores by Group (DCD)

	Rutter A (2) N = 107 Total Score 62 Mean (SD)	Rutter A (2) Under cut-off /over cut-off	Hyperactivity Rutter A (2) Total Score 6 Mean (SD)	Unsociability Rutter A (2) Total Score 6 Mean (SD)
Group 1 DCD Severe N = 44	23.93 (9.02)	5/39	4.00 (1.86)	2.95 (1.63)
Group 2 DCD Moderate N = 19	19.47 (8.57)	6/13	3.61 (1.82)	2.28 (1.95)
Group 3 No DCD N = 44	12.91 (9.69)	25/19	1.85 (2.20)	1.07 (1.40)

The hypothesis that those children with high scores on the ABC checklist would have high scores on the Rutter A (2) scales, a measure of common behaviour problems, was investigated using one-way ANOVA. This revealed homogeneous variances (Levene's $F = 0.43$, d.f. = 2,104 ns) and indicated significant differences among means ($F = 15.79$, d.f. = 2,104 $p < .001$). Inspection of the data reveals a trend of increasing means, with the mean for both Group 1 (Severe DCD) and Group 2 (Moderate DCD) above the cut-off score of

13 on the Rutter A (2), indicating clinical behaviour problems in these groups. Post hoc Scheffe tests revealed significant differences between mean scores for Groups 1(Severe DCD) and Group 3 (No DCD). There was also a significant difference between mean scores of Group 2 (Moderate DCD) and Group 3 (No DCD). There was no significant difference between the means of Group1 (Severe DCD) and Group 2 (Moderate DCD). The box-plot in Figure 3.1 shows the inter-quartile range of scores on the Rutter A (2) by group. It demonstrates the increasing median in line with increasing motor difficulties as defined by groups. The error bars indicate the maximum and minimum values in each group.

FIGURE 3.1: Rutter A (2) Scores by Group



To further investigate the hypothesis that there would be a relationship between high scores on the ABC checklist and behaviour, analysis of the data was carried out to investigate whether those children above the cut-off score of 13 on the Rutter A (2) would have high scores on the ABC Checklist using independent t-tests.

A score above the 13-point cut-off on the Rutter scales is indicative of clinical behaviour problems (Rutter, Tizard and Whitmore, 1970).

The pattern of scores on the four sections of the checklist was also analysed in this way.

The results can be seen in Table 3.4.

TABLE 3.4: ABC Checklist by Rutter A (2) score below /above cut-off score

	ABC Checklist Mean (SD)	Section 1 Mean (SD)	Section 2 Mean (SD)	Section 3 Mean (SD)	Section 4 Mean (SD)
Above Rutter A (2) cut-off of 13 N=68	64.19 (29.38)	14.39 (7.04)	15.06 (8.05)	16.70 (8.50)	17.85 (8.80)
Below Rutter A (2) cut-off of 13 N=35	23.91 (26.33)	5.42 (6.22)	4.21 (6.02)	6.79 (8.19)	7.45 (8.93)
	t(101)=6.82 p<.001	t(98)= 6.21 p<.001	t(98)=6.85 p<.001	t(98)=5.55 p<.001	t(98)=5.53 p<.001

Section 1 Child Stationary/Environment Stationary
Changing

Section 3 Child Stationary/Environment

Section 2 Child Moving /Environment Stable

Section 4 Child Moving/Environment Changing

As can be seen from Table 3.4 there are significant differences between the mean scores of the two groups on total ABC Checklist score and on each of the sections of the Checklist.

Inspection of the data indicates that the children who have scored above the cut-off on the Rutter A (2) have higher mean scores on the total ABC Checklist score, and on each section, than the group who have scored below the cut-off score. There is a pattern of increasing mean scores across the sections in line with the increasing difficulty of the tasks.

However the pattern of scores does not confirm the subsidiary question that there would be a relationship between high scores on the Rutter A (2) scales and specific patterns of motor difficulty.

3.2.1.1 Hyperactivity Score – Rutter A (2)

To investigate the hypothesis that children with high ABC checklist scores would be more likely to show difficulty with hyperactive behaviour as measured by the Rutter A (2) scales, one-way ANOVA was used to explore the mean Hyperactivity score on the Rutter scales in the three groups. The groups showed homogeneous variances (Levene’s F=1.52, d.f. =

2,99 ns) and a significant difference in mean hyperactivity score across the groups ($F=12.91$, d.f. = 2,99, $p<.001$). Inspection of the data in Table 3.3 reveals the mean scores for both Group 1 (Severe DCD) (4.00) and Group 2 (Moderate DCD) (3.61) are above the cut-off score when hyperactivity is said to be present, indicating a positive relationship between the degree of motor difficulties and hyperactivity. Post Hoc Scheffe tests showed a significant difference between mean hyperactivity scores in Groups 1 (Severe DCD) and 2 (Moderate DCD) when compared separately to Group 3 (No DCD) but not between Groups 1 and 2.

3.2.1.2 Unsociability Score – Rutter A (2)

To test the hypothesis that children with high ABC checklist scores were more likely to have high scores on the Unsociability index of the Rutter A (2) scales a one-way ANOVA was used to look at the Unsociability score of the three groups. The groups show homogeneous variances (Levene's $F=1.96$, d.f. = 2, 99 ns) and significant variation in mean scores across the groups ($F=14.50$, d.f. 2, 99 $p<.001$). Inspection of the data in Table 3.3 reveals that although the mean scores of Group 1 (Severe DCD) (2.95) and Group 2 (Moderate DCD) (2.28) are significantly higher than the mean score of Group 3 (1.07) they do not reach the cut-off score of 3 when unsociability is said to be present. Post Hoc Scheffe tests indicate significant differences in the mean scores between Groups 1 (Severe DCD) and 2 (Moderate DCD), when compared separately to Group 3 (No DCD) but not between Group 1 and Group 2.

3.2.2 Adapted Autism Spectrum Screening Questionnaire (AASSQ)

The hypothesis that children with motor impairment would be more likely to have high scores on a measure of autistic like behaviour was tested by a series of analyses.

The hypothesis that children with high DCD scores, in particular those in Group1, (Severe DCD), and Group 2 (Moderate DCD) would differ significantly from children without DCD (Group 3), in their scores on the AASSQ (a measure of autistic like behaviour) was addressed using one way ANOVA. Specifically, one-way ANOVA was used to look at the differences among the AASSQ scores across the three groups. The results can be seen in Table 3.5.

TABLE 3.5: AASSQ Scores by Group

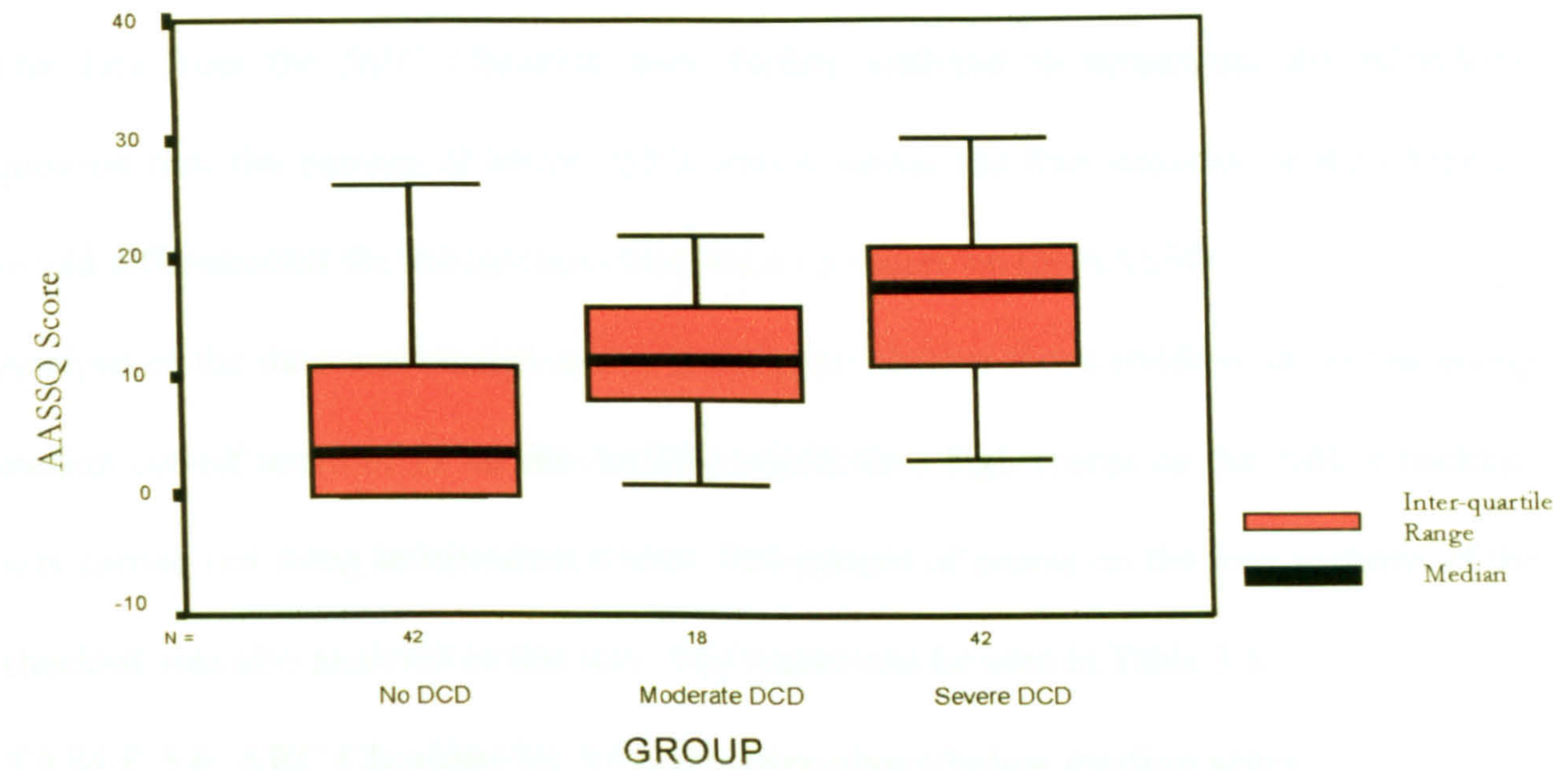
	AASSQ N=102 Total Score 30 Mean (SD)	AASSQ ^b Under cut-off /over cut-off
Group 1 DCD Severe N =44	16.60 (6.54)	9/33
Group 2 DCD Moderate N = 19	12.22 (7.40)	8/10
Group 3 No DCD N = 44	6.21 (6.78)	31/11

The groups showed homogenous variances (Levene’s $F = 0.20$ d.f. = 2,99 ns) and significant differences among mean scores across the groups ($F= 24.62$, d.f. = 2,99 $p<.001$). Inspection of the data in Table 3.5 reveals a pattern of increasing means. The mean scores for the groups increase in line with the degree of DCD. Post hoc Scheffe tests indicated Group 1 (Severe DCD) and Group 2 (Moderate DCD) have significantly higher mean scores on the AASSQ than Group 3, (No DCD) when compared separately. However, there is not a significant difference between mean AASSQ scores for Group 1 (Severe DCD) and Group 2(Moderate DCD).

The pattern of increasing median AASSQ scores in the three groups is shown in Figure 3.2. The box-plot in Figure 3.2 shows the inter-quartile range of scores on the AASSQ by group. It demonstrates the increasing median in line with increasing motor difficulties as

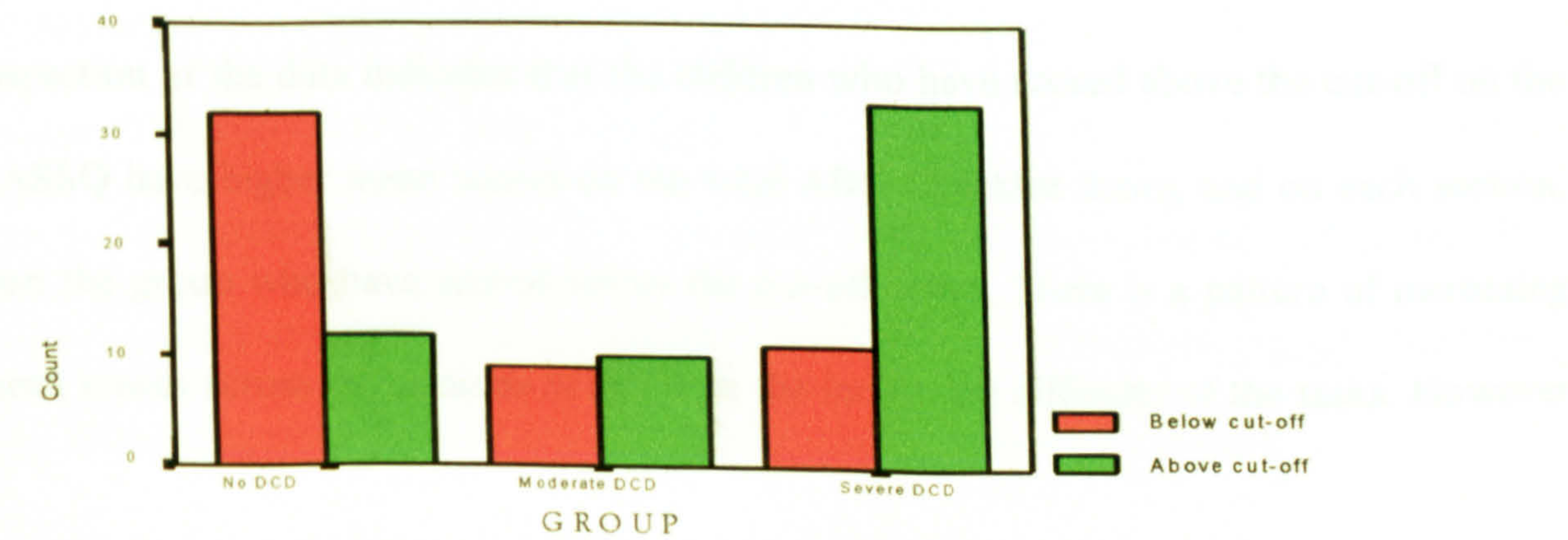
defined by groups. The error bars indicate the maximum and minimum values in each group.

FIGURE 3.2: AASSQ score by Group



To further investigate the hypothesis that the scores of children with higher ABC checklist scores would differ from those with low ABC checklist scores on the AASSQ a Chi square analysis was carried out. The results can be seen in Table 3.5. The median score of 11 on AASSQ for the whole group was used as the cut-off score. For each group the AASSQ scores were divided into those at the median score of eleven and above and those whose scores were below the median score. Figure 3.3 shows the distribution of scores above and below the median score of 11 across the three groups.

FIGURE 3.3: AASSQ Score above/below cut-off by Group



Analysis of the data shown in Figure 3.3 indicated a significant difference in the ratio of children above or below the median in Groups 1 (Severe DCD), 2 (Moderate DCD) and 3 (No DCD). ($\chi^2 = 23.19$ d.f. =2 $p < .001$).

The data from the ABC Checklist were further analysed to investigate the subsidiary question that the pattern of motor skills shown across the four sections of the checklist would differentiated the sub-groups of children by scores on the AASSQ.

Analysis of the data was carried out to investigate whether those children above the group median cut-off score of 11 on the AASSQ would have high scores on the ABC Checklist, was carried out using independent t-tests. The pattern of scores on the four sections of the checklist was also analysed in this way. The results can be seen in Table 3.6.

TABLE 3.6: ABC Checklist by AASSQ score above/below median score

	ABC Checklist Mean (SD)	Section 1 Mean (SD)	Section 2 Mean (SD)	Section 3 Mean (SD)	Section 4 Mean (SD)
Above AASSQ cut-off N=55	64.24 (29.64)	14.21 (7.04)	15.62 (7.83)	17.33 (8.64)	18.36 (8.71)
Below AASSQ cut-off N=52	35.04 (31.38)	8.81 (7.93)	7.12 (7.97)	9.13 (8.52)	9.98 (9.53)
	t(105)=-4.95 p<.001	t(102)=-3.67 p<.001	t(102)=-5.48 p<.001	t(102)=-4.86 p<.001	t(102)=-4.68 p<.001

Section 1 Child Stationary/Environment Stationary
Section 2 Child Moving /Environment Stable

Section 3 Child Stationary/Environment Changing
Section 4 Child Moving/Environment Changing

As can be seen from Table 3.6 there are significant differences between the mean scores of the two groups on total ABC Checklist score and on each of the sections of the Checklist. Inspection of the data indicates that the children who have scored above the cut-off on the AASSQ have higher mean scores on the total ABC Checklist score, and on each section, than the group who have scored below the cut-off score. There is a pattern of increasing mean scores across the sections in line with the increasing difficulty of the tasks. However

the results do not indicate any specific areas of difficulty for children with scores above or below the AASSQ cut-off score of 11.

3.3 Group analysis – DCD and behaviour

The analysis of the data so far has indicated links between both Rutter A (2) scores and AASSQ scores and ABC Checklist scores. To investigate the hypothesis that there would be differences across the groups allocated on basis of DCD and presence or otherwise of clinical behaviour problems, on the measure of autistic like behaviour (AASSQ), the children were re-allocated to groups on the basis of degree of DCD (based on cut-off criterion of 5th and 15th percentile) and presence or absence of behaviour problems as measured by the Rutter A (2) scales (scores above or below the cut off score of 13). The groups were organised as follows:

Group A: DCD/ Behaviour Problems

This group consisted of children identified by the 15th percentile cut-off on the ABC Checklist and a score of 13 and above on the Rutter A (2). Parents had reported motor problems as measured by the Checklist and clinical behaviour problems.

Group B: DCD/ No Behaviour Problems

This group consisted of children identified by the 15th percentile cut-off on the ABC Checklist, and a score of less than 13 on the Rutter A (2). The parents of these children reported motor problems, but the children are not reported to have problems with behaviour.

Group C: No DCD/Behaviour Problems

This group consisted of children not identified by the 15th percentile cut-off on the ABC Checklist, and a score of 13 or more on the Rutter A (2). The children are not reported to have motor difficulties but are reported to have clinical behaviour problems.

Group D: No DCD/No Behaviour Problems

This group consisted of children not identified by the 15th percentile cut-off on the ABC Checklist and a score of less than 13 on the Rutter A (2). No problems with motor skills or behaviour are reported in this group.

3.3.1 Motor impairment and Behaviour

A further analysis of the groups was carried out using one-way ANOVA to investigate the patterns of motor difficulties seen in the four groups. The mean scores for the four sections of the ABC Checklist can be seen in Table 3.7.

TABLE 3.7: ABC Checklist Scores and AASSQ scores by Group and Behaviour

	ABC Checklist Total score 144 Mean (SD)	AASSQ Score Total score 30 Mean (SD)
Group A (DCD/ behaviour problems) N=50	76.75 (20.80)	16.60 (6.57)
Group B (DCD /no behaviour problems) N=10	56.36 (16.88)	8.70 (5.68)
Group C (No DCD/ behaviour problems) N=19	26.74 (11.92)	11.11 (6.18)
Group D (No DCD /no behaviour problems) N=23	9.44 (12.78)	2.17 (4.09)

To investigate the hypothesis that there would be a difference among the four groups on ABC Checklist scores, a one-way ANOVA was carried out on the data in Table 3.7. This revealed significant variances in the means (Levene’s $F= 5.28$, d.f. =3, 103, $p<.002$) and significant differences among mean total ABC Checklist scores ($F=97.42$, d.f. = 3,103 $p<.001$). As the group sizes are different and population variances are different, following guidelines from Field (2000), Games-Howell post hoc tests were used. Post hoc tests showed significant differences between each group when compared separately with the other three groups. Inspection of the data indicates a trend of increasing means across the groups.

3.3.2 AASSQ by Group (DCD and Behaviour)

The hypothesis that there would be differences across the groups on the measure of behaviour characteristic of Asperger syndrome (AASSQ) was investigated using one-way ANOVA. The mean AASSQ scores for the four groups are shown in Table 3.7.

One-way ANOVA revealed significantly different variances in the means across the groups, (Levene's $F = 3.19$ d.f. 3, 98 $p < .03$). One-way ANOVA indicated significant difference among mean AASSQ scores ($F=31.92$, d.f. = 3, 98 $p < .001$) between the groups. As the group sizes are different and population variances are different, following guidelines from Field (2000), Games-Howell post hoc tests were used. Games-Howell post hoc tests indicated significant differences in mean AASSQ scores between Group A (DCD, Behaviour problems), and the three other groups (DCD No Behaviour problems, and both groups with no DCD) when compared separately. There was not a significant difference between the mean AASSQ score of Group B (DCD No behaviour problems) and Group D (No DCD, Behaviour problems}. The mean AASSQ score of Group 4 (NO DCD, No Behaviour Problems) was significantly different from the three other groups when compared separately. Inspection of the data indicates the mean score for Group 4 is lower than the mean score for each of the three other groups. Inspection of the data also indicates that the mean AASSQ score for Group C (No DCD /Behaviour) is at the median cut-off point of 11 suggesting these children have some difficulties in this area although not as large as those who have both DCD and behaviour problems.

3.4 Relationships among Measures

The original three groups were formed using arbitrary cut-offs on the basis of ABC checklist scores. This arbitrary division into groups can cause loss of information about the nature of the relationships across the data. To overcome this, the data were also analysed

by the use of correlations and regression. The series of investigations carried out above comparing the three groups and the four groups has indicated relationships among high scores on the ABC Checklist (Motor difficulties), high Rutter A (2) scores (behaviour problems) and high AASSQ scores (Asperger’s behaviour).

3.4.1 ABC Checklist, Rutter A (2), AASSQ, and Age

To investigate the hypothesis that there is a correlation between age and motor difficulty, problem behaviour and autistic like behaviour, correlations were carried out. The norms for the ABC Checklist (Sugden & Henderson 1982) are age related and indicate a negative relationship between motor scores and age, i.e. lower scores are required to reach the cut-off points with increasing age. This relationship has been confirmed by the correlations between age and ABC Checklist. As can be seen in Table 3.8 there is a negative correlation between age and motor difficulty (ABC Checklist).

Table 3.8: Correlations among Age, ABC Checklist, Rutter A (2), Rutter A (2) Hyperactivity/ Unsociability, AASSQ for Total Group

	ABC Checklist	Rutter A (2)	Rutter A (2) Hyperactive	Rutter A (2) Unsociability	AASSQ
AGE	-.37** N=107	-.28** N=107	-.38** N=102	-.11 N=102	-.07 N=102
ABC Checklist		.68** N=107	.62** N=102	.54** N=102	.63** N=102
Rutter A (2)			.74** N=102	.79** N=102	.69** N=102
Rutter A (2) Hyperactive				.49** N=102	.47** N=101
Rutter A2 Unsociability					.67** N=101

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

There are also statistically significant correlations between age and behaviour problems (Rutter A (2) scores) and age and hyperactive behaviour indicating that scores on these three variables are likely to decrease with increasing age. There is no correlation between age and unsociability as measured by the Rutter A (2) or between age and AASSQ.

To further investigate the relationships between scores on the ABC Checklist, the Rutter A (2) scales (behaviour problems) and AASSQ (behaviour characteristic of Asperger syndrome) a series of correlations was carried out.

Analysis of the three variables, ABC checklist, Rutter A (2) and AASSQ indicates positive correlations between ABC Checklist and Rutter A (2) scores, (Pearson $r = 0.68$ $p < 0.01$), ABC Checklist and AASSQ (Pearson $r = 0.63$ $p < .001$), AASSQ and Rutter A (2) scores, (Pearson $r = 0.69$ $p < .001$). There is also a positive correlation between unsociability and AASSQ score (Pearson $r = .67$ $p < .001$), and a reduced correlation between hyperactivity and AASSQ (Pearson $r = .47$ $p < .001$).

As would be expected, because they are from the same scale, there are strong correlations between total Rutter A (2) score and Rutter Hyperactivity and Unsociability scores but not between Hyperactivity and Unsociability scores.

As a result of these positive correlations a further analysis was carried out to hold each variable constant to consider the difference each variable makes to the overall correlations. To further investigate the hypothesis that children with high scores on ABC checklist would have high scores on Rutter A (2) scores, independent of AASSQ, partial correlations were carried out between Rutter A (2) scores and ABC Checklist, controlling for AASSQ (Pearson Partial $r = 0.42$ $p < .001$) indicating a statistically significant correlation between Rutter A (2) scores and ABC Checklist scores.

To investigate the hypothesis that children with high ABC Checklist scores would have high AASSQ scores independent of Rutter A (2) scores, a further partial correlation, controlling for Rutter A (2) scores was carried out. The resulting partial correlation (Pearson Partial $r = 0.30$ $p < .002$) showed a statistically significant correlation between AASSQ scores and ABC Checklist scores. Neither of these correlations is as strong as when the three variables are included.

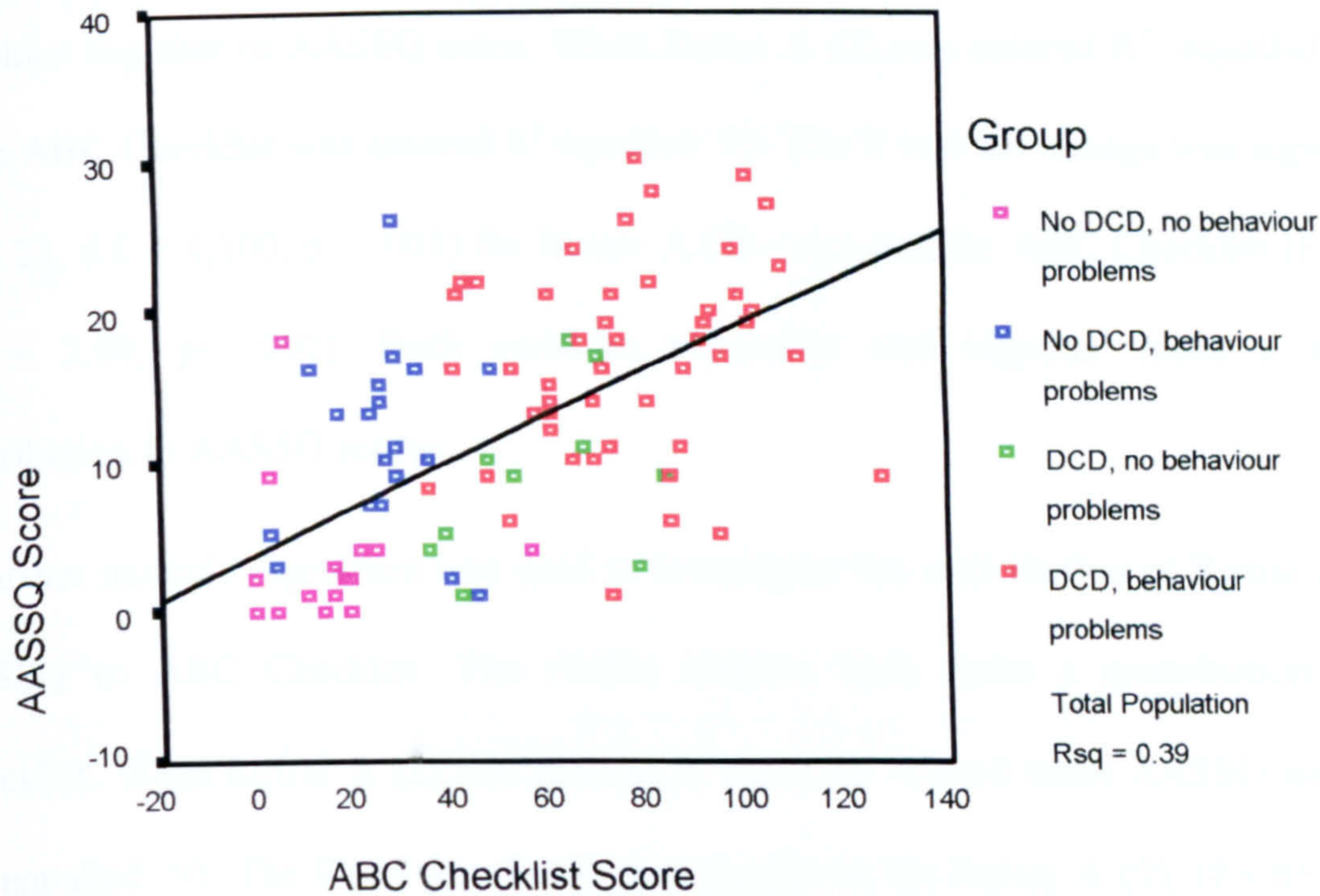
The hypothesis that children who had high scores on the Rutter A (2) scales would have high scores on the AASSQ, independent of motor skills, was tested using a partial correlation between Rutter A (2) scores and AASSQ scores, controlling for motor problems as measured by the ABC checklist. When the effects of motor skills are controlled for, there is a significant partial correlation between AASSQ score and Rutter A (2) scores (Pearson Partial $r = 0.46$ $p < .001$).

Correlations were also used to investigate the effect of age on motor scores and measures of behaviour. When controlling for age there remains a statistically significant correlation between ABC checklist and Rutter A (2) scores, (Pearson Partial $r = 0.65$, $p < .001$), ABC checklist and AASSQ scores (Pearson Partial $r = 0.64$ $p < .001$) and between AASSQ and Rutter A (2) scores (Pearson Partial $r = 0.70$ $p < .001$).

3.4.2 ABC Checklist, Rutter A (2) and AASSQ

As a further research question to confirm the indications from previous statistical analyses, a basic regression analysis was carried out to investigate the relationship between ABC Checklist scores and AASSQ scores with AASSQ as the dependant variable. Figure 3.4 shows the line of best fit for these two variables, with individual scores labelled by DCD and presence or absence of behaviour problems.

FIGURE 3.4: AASSQ by ABC Checklist



A linear regression with AASSQ as the dependent variable and ABC checklist held constant indicates that 39% of the variance in AASSQ is predicted by ABC checklist score. ($r^2 = .39$, $p < .001$). Inspection of the scattergram in Figure 3.4 indicates a high number of children in Group A (DCD, Behaviour Problems) have high scores on the AASSQ.

To further investigate the relationships among the three variables, ABC Checklist, Rutter A (2) and AASSQ, a multiple regression was carried out. The variables, AASSQ, and ABC checklist, were entered in sequence to assess the relationship with Rutter A (2). The results indicate AASSQ score makes a major contribution to Rutter A (2), ($R^2 = .48$), and when ABC checklist was entered ($R^2 = .57$). The F test of change was statistically significant when AASSQ was entered ($F=92.23$, d.f.1, 100, $p < .001$) and for ABC Checklist ($F = 66.03$, d.f. = 2, 99, $p < .001$). $R^2 = .57$ once AASSQ is followed by ABC checklist suggesting around 57% of the variance in Rutter A (2) scores is explained by the two variables, ABC Checklist and AASSQ score.

Multiple regression was also used to investigate the contribution of Rutter A (2) and ABC Checklist together to AASSQ score. When Rutter A (2) was entered R^2 equalled .48, and when ABC Checklist was entered R^2 equalled .52. The F test for change was significant ($F = 42.23$, d.f. = 1,100, $p < .001$) for Rutter A (2) score and for ABC Checklist ($F = 55.17$, d.f. = 2,99, $p < .002$). Both variables separately and together make a significant contribution to AASSQ scores.

A further multiple regression was used to investigate the contribution of Rutter A (2) and AASSQ to ABC Checklist. The results indicate both make a contribution to ABC Checklist. When Rutter A (2) was entered R^2 equalled .45 and when AASSQ was entered R^2 equalled .50. The F test for change was significant for Rutter A (2), ($F = 81.25$, d.f. = 1,100 $p < .001$) and for AASSQ ($F = 49.20$, d.f. = 2, 99, $p < .001$)

Overall the analyses indicate the three variables, ABC Checklist, Rutter A (2) and AASSQ scores together explain the variance in each other. Although these results do not suggest causal primacy they do indicate that ABC Checklist and Rutter A (2) scores together predict the presence of autistic like behaviour.

3.4.3 Relationships among measures in sub-group with IQ assessment

Finally a further analysis was carried out on the data from the group of children who had completed a full IQ assessment. As a subsidiary research question, correlations were carried out to investigate the relationship between ABC checklist scores and the Rutter A (2) measures and the AASSQ. The results can be seen in Table 3.9.

Table 3.9: Correlations among ABC Checklist, Rutter A (2), Hyperactivity/ Unsociability, AASSQ and IQ

	ABC Checklist	Rutter A (2)	AASSQ	Full Scale IQ	Rutter A(2) Hyperactivity	Rutter A(2) Unsociability
ABC Checklist		.63** N=38	.65** N=38	-.45** N=37	.50** N=37	.66** N=37
Rutter A(2)			.78** N=37	-.39* N=38	.71** N=37	.84** N=37
AASSQ				-.57** N=37	.50** N=37	.73** N=37
Full Scale IQ					-.50** N=37	-.21** N=37
Rutter A(2) Hyperactivity						.53** N=37

** Correlation significant at the .01 level (two tailed)
* Correlation significant at the .05 level (two tailed)

The results indicate a negative relationship between ABC Checklist score and Full Scale IQ, suggesting children with lower IQ scores are likely to have higher ABC checklist scores, indicating more difficulties with motor skills. There is also a statistically significant positive correlation between ABC Checklist score and Rutter A (2) scales and the AASSQ, and between ABC Checklist and Rutter Hyperactivity score and the Unsociability score. Unlike the total group there is a stronger positive correlation between ABC Checklist score and unsociability than hyperactivity. This may of course reflect the composition of children in this small group and is not representative of the whole group. The correlations between ABC Checklist, Rutter A (2), Hyperactivity, Unsociability and AASSQ when Full Scale IQ is controlled for, are reported in Table 3.10 below.

TABLE 3.10: Correlations among ABC Checklist, Rutter A (2), Hyperactivity, Unsociability, and AASSQ controlling for IQ.

	Rutter A (2)	Rutter A (2) Hyperactivity	Rutter A (2) Unsociability	AASSQ
ABC Checklist	.64** N=38	.50** N=37	.66** N=37	.65** N=37
Rutter A (2)		.71** N=37	.84** N=37	.77** N=37
Rutter A (2) Hyperactivity			.53** N=37	.50** N=37
Rutter A (2) Unsociability				.73** N=37

** Correlation significant at the .001 level (two tailed)

As can be seen from Table 3.10 the correlations between ABC Checklist, Rutter A (2) and AASSQ scores are still present. The results of the investigation of relationships between variables in the sub-group of children, who have had complete WISC 111 assessments, confirm findings from the total group, that there is a relationship between ABC Checklist scores and measures of both clinical behaviour problems and autistic behaviour measured by the AASSQ.

3.5 Summary of Results

The aim of the study was to investigate patterns of behaviour, including the presence of clinical behaviour problems, and autistic like behaviour shown by children with DCD and to see how these differed from the behaviour seen in children without DCD. The main question investigated proposed that children who have severe problems in motor skills, defined by the cut-off criterion of 15th percentile on the ABC checklist (Henderson & Sugden 1992), would also show behaviour problems and difficulties with autistic like behaviour. The children in the study (aged six to twelve years) were allocated to three groups based on the degree of DCD. There were no significant differences in mean age across the groups. Although there were more boys than girls in each group this is consistent with referrals to developmental services.

A full WISC-111^{UK} was completed on a sub-group of thirty-eight children and the mean Full Scale IQ was within the normal range.

The results of this investigation have indicated that there is a positive relationship between presence and degree of DCD, clinical behaviour problems and autistic like behaviour. Those children with the most severe DCD showed the highest level of clinical behaviour problems and autistic like behaviour. There was a pattern of increasing means across the three groups indicating a relationship between behaviour problems and increasing degree of motor impairment.

Results have also indicated significant differences in the mean Rutter Scale hyperactivity and unsociability scores. The mean score for both Group 1 (Severe DCD) and Group 2 (Moderate DCD) are above the cut-off score where hyperactivity is said to be present, but the scores for the two groups did not reach the cut-off point where unsociability is said to be present. Children in the Severe DCD group had the highest mean scores on both hyperactivity and unsociability.

Those children with scores above the cut-off of 13 for the Rutter A (2) scales have significantly higher mean ABC Checklist scores than those with Rutter A (2) scores below the cut-off. Similarly those children with AASSQ scores above the cut-off score of 11 had significantly higher mean ABC Checklist scores than those with AASSQ scores below the cut-off.

When grouped according to presence or absence of DCD together with presence or absence of behaviour problems (Rutter A (2) scores), the mean AASSQ shows a pattern of increasing means across the four groups, with the mean score for each group being significantly different from each of the other three groups. Group A (DCD, behaviour problems) had a significantly higher mean AASSQ score than the three other groups.

Further analyses to investigate relationships among measures indicated a significant positive correlation between ABC Checklist and Rutter A (2) scores, and between ABC Checklist and AASSQ score. There is also a significant positive correlation between Rutter A (2) scores and AASSQ scores.

There is a significant correlation between unsociability index and AASSQ, which is higher than the correlation between hyperactivity and AASSQ.

A further analysis was carried out on the data from the total group of children who had completed a full IQ assessment. Correlations were carried out to investigate the relationship between ABC checklist scores and the Rutter A (2) measures and the AASSQ. The results indicate a modest negative relationship between ABC Checklist score and Full Scale IQ, suggesting children with lower IQ scores are likely to have higher ABC checklist scores. There was also a positive correlation between ABC Checklist score and both Rutter A (2) scales and the AASSQ, and between ABC Checklist and Rutter Hyperactivity score and the Unsociability score, as with the findings from the total group.

CHAPTER 4

4.1 Discussion

The aim of this study was to investigate the patterns of behavioural difficulties seen in children with developmental coordination difficulties. A further aim was to investigate whether children with developmental coordination difficulties show an increased incidence of autistic like behaviour. Although a review of the literature indicates studies that have looked at motor impairment in children with autistic spectrum disorders, including Asperger syndrome, and other studies have documented difficulties with behaviour and attention/hyperactive behaviour in children with motor impairment, there have been no studies, which have aimed to look at the presence of autistic behaviour in children with motor impairment. Oppositional defiant disorder and hyperactivity are both frequently described in the literature as a feature of behaviour in children with motor impairment, but no one has investigated whether these patterns of difficulty are a feature of a wider diagnosis of autistic spectrum disorder.

The results of the present study have supported the main question. They confirm that for a group of children with DCD there is a greater likelihood that they will show an increased incidence of clinical behaviour problems and these are likely to be autistic like the patterns of behaviour. This relationship was investigated in a variety of ways and established that both the presence of DCD and degree of DCD are associated with clinical behaviour problems and increased behavioural difficulty characteristic of autistic spectrum disorder. Children with high scores on the AASSQ are also more likely to have scores on the ABC Checklist, which fall within the severe DCD group.

The data also showed a correlation between hyperactive behaviour and motor impairment/DCD. Gillberg and colleagues (1989, 1991) have reported this association but

the association observed in the present population is not as strong as that between DCD and autistic like behaviour.

An investigation by Kadesjo and Gillberg (1999) into the incidence of DCD in a total population explored the incidence of Asperger syndrome within the DCD population. However Kadesjo and Gillberg (1999) did not screen the total group for Asperger syndrome, only investigating those children who were reported to show difficulties with social interaction. They found a low rate of Asperger syndrome but they and other studies (Pine, Wassermann, Fried, Parides & Shaffer, 1997) found, as did the present study, a relationship between high levels of motor impairment and increased difficulties with behaviour, be it attention/hyperactivity, anxiety etc. More recent studies, (Miller and Ozonoff, 2000; Green et al, 2002) found a relationship between increased motor impairment and Asperger syndrome but both these studies were published after the present study and again are looking at more detailed aspects of motor impairment in Asperger syndrome rather than autistic spectrum disorder in children with DCD. Miller and Ozonoff (2000) indicated that motor impairment did not differentiate between Asperger syndrome and high functioning autism.

The strongest relationships observed in the present study were for interactions among the three variables.

As this study is based on parent report it is important to consider whether parents provide reliable information regarding their child's behaviour and development. Parental measures may affect the results in terms of rater bias, and self-selection and may have inflated the group correlations between measures. However, several studies, including one by Glascoe and Sandler (1995) encompassing 737 children, have shown that parental concerns about speech and language development, behaviour and other developmental issues were highly

sensitive (i.e. 75% to 83%) and specific (79% to 81%) in detecting global developmental deficits (Glascoe & Sandler 1995). The study by Glascoe and Dworkin (1995) combined parental concern with standardised parental report and found this combination to be effective for early behavioural and developmental screening in primary care settings. In a review of the status of the Rutter Parent and Teachers' scales, Rutter and Elander (1996) refer to the fact that ratings from parents and teachers may vary considerably, due in part to differences in the child's behaviour at home and at school. The scales were developed to provide identification of broadly defined but clinical, behavioural disturbance and are reported to work best when behavioural changes or patterns are well established over long periods. Rutter and Elander (1996) conclude that parent ratings may be more appropriate when the children under investigation are attending different schools, particularly different types of schools and where the research questions are looking at relationships between disturbed behaviours and other research questions. The present study fulfils these criteria and although the information provided is based only on parent report, it is likely to have some validity as it has used standardised measures. However parental bias may have influenced the results more than might be expected because the study was based in a tertiary referral centre. It is also important to consider whether the selection of participants, which included a group of participants referred to the tertiary service, has introduced a bias into the results thus making it more likely that children will present with behaviour problems, particularly behaviour characteristic of autistic spectrum disorder. An attempt was made to reduce referral bias by recruiting children through the Dyspraxia Foundation and ensuring that there was a representative sample of typical children; fifty per cent of the children have been recruited through the Neurodevelopmental Assessment Clinic, fifty per cent are self-referrals, (including those recruited through the Dyspraxia

foundation). The sample group of children may not reflect the general population, including those with a diagnosis of Developmental Coordination Disorder. Parents may have been influenced to take part by the level of concern they had about their child's particular set of difficulties, including concerns about behaviour. They may also have been influenced to agree to take part in the study because it was based in a tertiary referral centre resulting in an artificially high level of reported difficulties with clinical behaviour problems in the study population. However the literature indicates a high level of associated behaviour disorder in children with a diagnosis of Developmental Coordination Disorder, prevalence rates for DCD being suggested as 5-10% of population, with more boys than girls being affected. The male to female prevalence rates in the group reflect that seen in referrals to Developmental Assessment Clinics, and reflect the rate of DCD diagnosed in boys and girls. Gillberg and Gillberg (1989) reported up to 89% of children with DCD have associated behavioural difficulties. In the present study population the prevalence rates for scores above the cut-off on the Rutter A (2) scales in children with DCD is 82%, similar therefore to rates for the presence of clinical behavioural difficulties in children with DCD that has been reported in the literature. In addition the present study has found 68% of children within the DCD group have score over the group median score of 11 on the AASSQ, and when the population is further broken down into groups according to degree of DCD, 75% of those selected by the 5th percentile cut-off on the ABC Checklist had scores above the median score of eleven, and 53% of those at the 15th percentile cut-off on the ABC Checklist had scores above the median cut-off of eleven on the AASSQ. Only 27% of the group with no DCD had scores above the cut-off on the AASSQ. The children with DCD are therefore showing a high level of autistic like behaviour. It is possible that these high scores represent a high level of misdiagnosis in the

DCD group and that many of the children have since received a diagnosis of autistic spectrum disorder.

It is important to consider behavioural difficulties within the framework of the child's developmental level or cognitive profile. Intellectual status is an important consideration in understanding the context of child's motor difficulties, skills in social interaction and the presence of clinical behaviour problems. Although child's level of cognitive functioning is not required to be in the normal range for a diagnosis of DCD the child's motor difficulties are compared against age appropriate motor functioning. Ideally the results would have been supported by full IQ assessments on the total group. Due to time and practical constraints the sample of children in the study who have had full cognitive assessments is small. Thirty two percent of children in the DCD group completed a full IQ assessment and forty-one percent of the non-DCD group. The mean for the whole group is within the average range, suggesting that as a group they are representative of the general population of children with cognitive skills in the normal range. It is likely therefore that any behavioural difficulties described are independent of the child's intellectual status and in this sub group of children the results confirm that when IQ is controlled for there are strong correlations between motor skills, clinical behaviour problems and autistic like behaviour. The present study confirms the association between degree of motor difficulty and degree of behavioural difficulty, particularly autistic like behaviour, but finds a stronger association than previously reported. During the time the present investigation has been carried out there has been further discussion in the literature about diagnostic criteria for DCD (Dewey, Kaplan, Crawford & Wilson 2002) but no studies have further investigated associated patterns of behaviour or the presence of autistic like behaviour. Studies have begun to report an association between DCD, social problems and difficulties with peer

relationships. The DSM-1V criteria specifically exclude those children with a primary diagnosis of pervasive developmental disorders but although children were included in the study if they had a primary concern around motor impairment, the results suggest that these motor difficulties often co-occur with behavioural difficulties or other diagnostic categories. Although the diagnosis of an Autistic Spectrum Disorder in individual children cannot be confirmed, the results from the AASSQ indicate that further investigation of these children to consider such a diagnosis would be appropriate given the number of children reported to be showing autistic like behaviour. As mentioned, of the children in the sample with scores fulfilling criteria for DCD (based on motor skills alone), 68% scored above the median score on the AASSQ. Hellgren et al (1994) found a rate of 23% of the children who had severe motor control/perception dysfunction and Attention Deficit Disorder diagnosed at age seven fulfilled criteria for a diagnosis of Asperger syndrome. It has been noted in more recent studies (Green, Baird, Barnett, Henderson, Huber, & Henderson 2002) that when a sample is taken from a larger more typical population of children referred to paediatric occupational therapy departments, there are few children without associated difficulties. Green et al (2002) found that out of fifteen children who fulfilled the initial criteria for inclusion in a group of children with DCD, (based on scores on the ABC) three were excluded because they met Autistic Diagnostic Interview-Revised criteria (Lord, Rutter, & Le Couteur, 1994). This suggests an incidence rate of 20% among the DCD population. Green et al (2002) also found that within their Asperger syndrome group the scores of all eleven children fell below the 15th percentile and nine of these were below the 5th percentile on a measure of motor impairment, indicating an association between Asperger syndrome and DCD. In the present study, the higher rate of children

above the group median cut-off on the AASSQ may reflect a selection bias of increased numbers of children with DCD and difficulties with behaviour.

The results also indicate that children with DCD are significantly more likely to have a autistic like behaviour and further investigation of this aspect of their development is strongly recommended.

Since the present research was developed there has been a growing awareness of the overlap between different developmental syndromes. There have been further attempts to define the criteria to describe DCD and it has been suggested (Dewey & Wilson 2001; Gillberg 2003; Henderson & Henderson 2003;) that the use of diagnostic criteria proposed by DSM IV allows further research to investigate DCD when it occurs alone and the extent to which it is co-morbid with other developmental disorders. The DSM-IV criteria specifically exclude those children with other pervasive developmental disorders. However, the present research attempted to distinguish between those children with and without behavioural difficulties, and had difficulty recruiting children with motor impairment but no associated behavioural confirming the findings in the literature reporting a co-occurrence of motor impairment and behavioural concerns.

It is possible that these results are highlighting a disorder in empathy as described by Gillberg (1992) and that disorders linked with motor impairment are common to a range of developmental disorders, such as Tourette's syndrome, obsessive compulsive disorder, semantic pragmatic disorder and ADHD, and do not require a specific diagnosis of Autistic spectrum disorder.

There is also the possibility that the link between autistic like behaviour and DCD is coincidental. Difficulties with social interaction are commonly reported in children who have had some form of brain insult, either as a result of a medical condition or an acquired

brain injury. For example children with hydrocephalus, (Fernell, Gillberg, & von Wendt, 1991, 1991) immunodeficiency (Rogers, Lewin, Fairbanks, Geritsen & Gaspar 2001) heart conditions, stroke, epilepsy (Caplan Guthrie Komo Siddarth et al 2002) neurological damage, or low birth weight have all been described as displaying difficulties with peer relationships and social interaction. These children are also likely to have difficulties with motor skills.

Also of interest are the results of the investigations of the sub-group with the children who had full IQ assessments carried out. The results indicated a correlation between children with lower IQ, increased motor difficulties, behavioural difficulties and autistic like behaviour. These results may reflect the patterns of difficulties in children with developmental difficulties, although it is of note that the children were functioning within the normal range, IQ of 70 and above.

Although the present study may not be representative of a total population of children with DCD, studying this population highlights the links between behavioural difficulties, autistic like behaviour and DCD. It is likely that the clinic sample includes more severe cases and more extreme examples of the association between DCD and autistic behaviour. Perhaps the children in the present study over represent cases with comorbidity but the results do not contradict previous research but rather confirm the association.

4.2 Clinical Implications

The results of the present study indicate a strong relationship between motor impairment, behavioural difficulties and autistic like behaviour. The data support the need for careful assessment of behaviour in children with a diagnosis of Developmental Coordination Disorder particularly those with more severe motor difficulties. It also supports the need for careful assessment of motor skills in children with a diagnosis of autistic spectrum

disorder. As the literature reports long term implications for children with DCD and autistic spectrum disorder, early identification of specific difficulties and the links between these two diagnostic categories can lead to the development and maintenance of ongoing treatment and educational support programmes. Anecdotal evidence from clinical work has drawn attention to difficulties many children with DCD have with social interaction and peer relationships and it has been observed that these difficulties remain even when the child has learnt to compensate for many aspects of their motor impairment. As the children mature it is the impact of their social and communication difficulties that are of concern to parents and make the children more vulnerable, particularly in an educational setting. This study has highlighted the association between DCD and behavioural difficulties characteristic of children with a diagnosis of autistic spectrum disorder. The children are therefore likely to have long-term needs in promoting independence and self esteem. The present study has investigated a group of children aged between six and twelve years when social and behavioural difficulties are becoming more apparent.

Early diagnosis can prevent escalation of problems when the child's needs are not met particularly as social, academic and organisational demands increase when the child moves into the more challenging experience of secondary school.

Pro-active support programmes both in school and in the home may help to alleviate long-term difficulties. Parental report has indicated that difficulties with behaviour are likely to be pervasive. Although the data does not inform about causal relationships between DCD and autistic spectrum disorder, it does suggest that children with DCD may be at increased risk of autistic spectrum disorder and that this link is evident between the ages of six and twelve. Parents and teachers need to be alert to this association when considering the child's social, emotional and educational needs.

4.3 Limitations of study

The sample cannot be said to be representative of the total population of children with Developmental Coordination Disorder and there is the possibility of bias towards children with behavioural difficulties being included, reflecting the level of parents concern. However the study does provide a description of children with DCD and the patterns of behaviour and that can be associated with that diagnostic category.

Many recent studies investigating behaviour and social adaptation in children have used the Strength and Difficulties questionnaire (Goodman 1998) but this questionnaire had not been standardised on as wide a population as the Rutter A (2) when the present study began. Although the use of the Strength and Difficulties questionnaire would have made it possible to look in more detail at socialisation and relate the results to more recent studies, the scale has been validated against the Rutter scales, hence the scales are reporting on the same behaviours.

The AASSQ provides data about behaviour characteristic of autistic spectrum disorder, but analysis and interpretation of the data would be more straightforward if the complete ASSQ scale developed by Gillberg had been used. The scale has been developed further since data collection began on the present study and other scales have also been developed which may have been more suitable for the present study.

A questionnaire on the child's developmental history and present behaviour concerns was developed as part of the research, but this has not been analysed as there were difficulties with interpreting parent responses. Parent's recollection of events may be coloured by the child's subsequent development and their interpretation of the questions was not clear. The results would have been more relevant if the child's developmental history had been carried

out by parent interview rather than questionnaire and questions more r specifically directed to testing out the main hypotheses.

Many studies on children with DCD do not include full assessments of cognitive functioning and a complete assessment of all children in the study would have clarified the relationship between cognitive skills, and degree of motor impairment and behavioural difficulties.

4.4 Conclusions

This research project has investigated patterns of behaviour difficulty seen in children with a diagnosis of Developmental Coordination Disorder, giving particular consideration to the presence of autistic like behaviour. The investigation was carried out by parental report, using standardised questionnaires. The results confirm an association between increasing severity of motor difficulty and behavioural difficulties, including those behaviours characteristic of autistic spectrum disorder. Early investigation of the presence of behavioural difficulties in children with developmental coordination difficulties and investigation of the degree of motor impairment in children with a diagnosis of autistic spectrum disorder is recommended.

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APPENDIX

Great Ormond Street Hospital
for Children NHS Trust
and the Institute of Child Health
(University College London Medical School)



27 October, 1997

Ms J Gumley
Clinical Psychologist
Neurosciences Unit
Wolfson Centre
ICH

30 Guilford Street
London WC1N 1EH

Telephone: 0171 242 9789
Direct Fax: 0171 813 8234

Dear Ms Gumley

97NR19 Cognitive, social and behaviour profiles in clumsy children.

Notification of ethical approval

The above research has been given ethical approval after review by the Great Ormond Street Hospital for Sick Children NHS Trust / Institute of Child Health Research Ethics Committee subject to the following conditions.

1. Your research must commence within twelve months of the date of this letter and ethical approval is given for a period of 18 months from the commencement of the project. If you wish to start the research more than twelve months from the date of this letter or extend the duration of your approval you should seek Chairman's approval.
2. You must seek Chairman's approval for of proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature, ie. using the same procedure(s) or medicinal product(s). Each research project is reviewed separately and if there are significant changes to the research protocol, for example in response to a grant giving bodies requirements you should seek confirmation of continued ethical approval.
3. It is your responsibility to notify the Committee immediately of any information which would raise questions about the safety and continued conduct of the research.
4. Specific conditions pertaining to the approval of this project are:
 - The use of the enclosed standard consent forms for the research. A copy of the signed form must be placed in the patient's clinical records and a copy must be kept by you with the research records as our insurers may demand access to them.

Research and Development Office

Patron
Her Majesty The Queen

President
*Her Royal Highness
The Princess of Wales*

Chairman
Sir Brian Hill MA FRCS FRCR

Chief Executive
Robert Creighton MA

Yours sincerely

A handwritten signature in black ink that reads "Anna Jenkins". The script is cursive and fluid, with the first name "Anna" and last name "Jenkins" clearly legible.

Anna Jenkins
Secretary to the Research Ethics Committee

encs

cc Dr V Larcher

**Great Ormond Street Hospital for Children NHS Trust and
Institute of Child Health Research Ethics Committee**

**Consent Form for PARENTS OR GUARDIANS
of Children Participating in Research Studies**

97NR19 Cognitive social and behaviour profiles in clumsy children: Ms D Gumley

NOTES FOR PARENTS OR GUARDIANS:

1. Your child has been asked to take part in a research study. The person organising that study is responsible for explaining the project to you before you give consent.
2. Please ask the researcher any questions you may have about this project, before you decide whether you wish to participate.
3. If you decide, now or at any other stage, that you do not wish your child to participate in the research project, that is entirely your right, and if your child is a patient it will not in any way prejudice any present or future treatment.
4. You will be given an information sheet, which describes the research project. This information sheet is for you to keep and refer to. *Please read it carefully.*
5. If you have any complaints about the way in which this research project has been, or is being conducted, please, in the first instance, discuss them with the researcher. If the problems are not resolved or you wish to comment in any other way, please contact the Chairman of the Research Ethics Committee, by post via The Research and Development Office, Institute of Child Health, 30 Guilford Street, London, WC1N 1EH or if urgent, by telephone on 0207 242 9789 ex 2620 and the committee administration will put you in contact with him.

CONSENT

I/We _____, being the parent(s)/guardian(s) of

_____ agree that the Research Project named above has been explained to me to my/our satisfaction, and I/We give permission for our child to take part in this study. I /We give permission for our child to take part in this study. I/We have read both the notes written above and the Information Sheet provided and

understand what the research study involves.

SIGNED (Parent(s)/Guardian(s))

SIGNED (Researcher)

AASSQ BEHAVIOUR QUESTIONNAIRE

I would be helpful if you could make an assessment as to whether your child stands out as *different* from other children of his/her age.

	NO	SOMEWHAT	YES
1. Is old fashioned or precocious.			
2. Is regarded as an 'eccentric professor' by other children.			
3. Lives somewhat in a world of his/her own with restricted idiosyncratic intellectual interests.			
4. Accumulates facts on certain subjects (good rote memory) but does not really understand the meaning.			
5. Has a literal understanding of puzzling and metaphorical language.			
6. Has a different style of communication with formal fussy, old fashioned or 'robot-like' language.			
7. Has a different voice or speech.			
8. Is surprisingly good at some things and surprisingly poor at others.			
9. Uses language freely but fails to make adjustment to fit social contexts or the needs of different listeners.			
10. Lacks empathy.			
11. Makes naïve and embarrassing remarks.			

	NO	Somewhat	YES
12.Wishes to be sociable but fails to make relationships with peers.			
13. Lacks common sense.			
14. Is poor at games: no idea of cooperating in a team, scores ‘own goals’.			
15. Has special routines: insists on no change			

Are there any other reasons, which make this child, seem *different* from other children or the same age?

SECTION C

THE IMPACT OF DIAGNOSIS AND TREATMENT FOR A BRAIN TUMOUR ON SUBSEQUENT SOCIAL, EMOTIONAL AND COGNITIVE DEVELOPMENT OF A YOUNG GIRL

Introduction

This case has been chosen to demonstrate the extent to which late effects following diagnosis and treatment for a brain tumour can influence both the child and family's continuing adjustment to the illness. It also illustrates how taking a multi-dimensional approach, which is both dynamic and proactive, can inform recommendations and intervention to facilitate social and emotional adjustment and strategies for coping with the functional impact of the illness.

Background

The literature has reported long-term effects from diagnosis and treatment for brain tumours particularly where adjuvant therapy has included cranial radiotherapy (Hoppe-Hirsch et al., 1990; Jannoun & Bloom 1990). Less is known about the late effects on cognitive and emotional functioning of children who have had surgery only for tumours in the cerebellum, although recently more consequences have been documented (Hoppe-Hirsch et al., 1995; Kao et al., 1994; Ater et al., 1996). The children are usually reported to have IQ scores within the normal range (Palmer et al., 2001), but they have also been reported to exhibit slowed motor output and processing speed (Carpentieri et al., 2003), poor expressive language skills, poor attention and language sequencing, diminished verbal memory and some compromise of visuo-spatial processing and regulation of affect (Levisohn, Cronin-Golomb & Schmahmann 2000; Riva & Giorgi 2000).

Visual spatial difficulties are characterized by impairments in planning and organisational aspects of tasks. Deficits in expressive language are characterized by brief responses, lack of elaboration, reluctance to engage in conversation, long response latencies and word finding difficulties. These deficits can be quite subtle in their expression and are easily overlooked when considering a child's cognitive and emotional functioning.

Although the prognosis for tumours such as cerebella astrocytomas has improved from 40% to more than 90% (Magnani et al 2001), late effects will depend to some extent on invasiveness, and location of the tumour, on rate of growth and how destructive it has been to neural tissue.

Cerebella mutism following surgery in the posterior fossa has been reported in adults and children and has been described as a distinct clinical syndrome. The features include a postoperative period ranging from 18-72 hours when the child can speak, followed by a period of mutism, which cannot be explained by postoperative deficits of the cranial nerves or speech organs. The mutism is transient with recovery varying from weeks to months. Emotional lability is also often described during this period together with facial weakness and nystagmus. Children tend to go through a phase when they gradually regain speech and are often described as apathetic with some showing non-verbal reactions and some comprehension, whilst others do not. Although acute symptoms resolve, studies have indicated that these children are at increased risk of neuropsychological effects (Rey-Casserly, et al., 1997; Catsman-Berrevoets, et al., 1999).

Referral

Heather* (name has been changed to protect anonymity) was referred, aged 9 years 6 months, for a baseline assessment of her neuropsychological functioning. Guidelines for the treatment of children diagnosed with a brain tumour are reviewed and provided by the United Kingdom Children's Cancer Study Group and the referral was part of routine treatment protocol. Although no longer standard treatment, monitoring of neuropsychological profiles is a feature of a proactive service offered to families to anticipate and guide future intervention.

Initial Presentation

Heather was interviewed with her mother to gather information about the history of her illness and any concerns the family had about her ongoing cognitive, social and emotional development. As posttraumatic stress symptoms have been reported in families of children diagnosed with cancer (Kazak et al., 1998), the assessment began by asking Heather and her mother about the experience of the illness. They were very keen to tell their story, beginning with mother's attempt to understand and explain Heather's symptoms before diagnosis. A period of twelve months, during which repeated visits were made to the general practitioner, eventually culminated in the diagnosis of a brain tumour followed by immediate hospitalisation and surgery.

Heather lives at home with her mother and her younger brother. They have no contact with her father. Prior to the illness there had been no concerns about Heather's social and emotional functioning and she was reported to be making very good progress with schoolwork.

When Heather was eight years old she began complaining of feeling tired and pain in her head. She became progressively worse over a period of twelve months and three months prior to the initial interview, at the age of nine years, Heather was diagnosed with a low-grade cerebellar pilocytic astrocytoma in the posterior fossa. She subsequently underwent surgery and a complete macroscopic excision was achieved. She did not have radiotherapy or chemotherapy and was given a good long-term prognosis.

Following surgery Heather suffered cerebella mutism. As a result she was unable to speak and had difficulties with swallowing. She had help from speech and language therapy and her speech gradually improved although it was still described as 'rather quiet' six months later. At the time of the first assessment she was fluent. Heather also described significant

visual difficulties, including double vision and reported she was unable to see for a period following surgery. She had difficulties with mobility and was ataxic on the left side. Her symptoms are consistent with those described in the literature on cerebella mutism (Riva & Giorgi 2000; Levisohn et al., 2000). Both Heather and her mother describe this period in hospital as very distressing. Mother vividly describes the moment when Heather said her first word following the mutism and how the family worked to help her begin to speak again.

On leaving hospital Heather used a wheel chair, and although at the time of the assessment she was still using the wheelchair she no longer needed it full time. She still had left sided weakness and described herself as wobbly with a tendency to fall over and knock into things easily. She continued to get very tired. She was attending her local primary school two full days and most afternoons each week and although there were problems with writing skills and fine motor skills there were no reported problems around schooling or education. Headaches were a problem but not preventing her from attending school.

Provisional Hypothesis

At the time of this assessment Heather and her mother were still very shocked and distressed by the experience of diagnosis of a life threatening illness. They were pleased with the good prognosis, and with the progress Heather was making in all areas since diagnosis. They were coping well and although Heather was only attending school on a part-time basis they were optimistic about her continuing progress. The school was providing a high level of support. The family were still in the early stages of adjusting and coping with the illness, but did not request any emotional support at this stage.

First Neuropsychological Assessment

To provide a baseline level of functioning in accordance with protocol, Heather was given

a neuropsychological assessment three months following surgery. Common difficulties for children with CNS tumours include memory, attention and learning difficulties, slowed speed of processing, or executive function impairments. Behavioural difficulties can include tiredness, irritability, and adjustment problems all of which could impact on the Heather's functioning within the school situation. The assessment was based on the model described by Rourke (1989) who suggests that relevant factors for consideration for children with CNS tumours should include:

- The types and number of skills impaired
- The degree of impairment
- Capacity for adaptation
- Quality of intact abilities.
- Social and behavioural description.

It was anticipated that there might be changes in cognitive functioning in line with literature on children who had had surgery as the only form of treatment following diagnosis for a brain tumour (Steinlin et al., 2003). The assessment aimed to describe these with a view to making recommendations for support, particularly in the educational setting.

The WISC 111 (Wechsler 1992) provided an assessment of cognitive skills, and literacy and numerical skills were assessed using the WORD (Wechsler 1993) and the WOND (Wechsler 1993). As a result of the assessment, Heather was reported to be functioning overall within the high range for her age. She had above average verbal comprehension and perceptual organisation skills.

Strengths on the Freedom from Distractibility index indicated good attention. Her scores on the Processing Speed index were weaker than other areas. Literacy skills were within the average range and mathematical skills were above average. A brief assessment of

memory was carried out which indicated a relative weakness on verbal memory. These results are as expected from the literature; overall cognitive functioning within the average range but some specific difficulties with slower processing speed and memory. There were no indications of any specific effects on language skills although difficulties with verbal memory were highlighted.

Two hypotheses were developed.

- As the literature has indicated that substantial parent and child distress can continue after treatment ends, and that post traumatic stress symptoms are higher in mothers of survivors of childhood cancer (Kazak et al., 1998) it was hypothesised that both mother's and Heather's anxiety may increase with time from treatment. A review was therefore recommended 18 months after this initial assessment, with provision for the family to contact the psychology service at any time if they had concerns.
 - It was also hypothesised that difficulty with verbal memory, visuo-motor skills, and processing speed may impact on Heathers continuing educational progress.
- Recommendations were made to assist developing memory skills.

Follow-up Assessment

At the age of 12 years 5 months, that is three years after the initial assessment, the Consultant Neurologist referred Heather for a follow-up assessment of her neuropsychological functioning. This referral followed Heather's admission to hospital to investigate continuing severe headaches. The family had not made contact with the psychology service in the meantime.

Previous Assessments

An assessment of Heather's emotional status was carried out during her stay in hospital prior to the second assessment. This included the Children's Headache Assessment Scale,

The Children's Health Locus of Control Scale (CHLC) The Birleson Depression Scale and the Spence Children's Anxiety Scale (SCAS). The results indicated the headaches were having a significant impact on Heather's general functioning and frequently disrupted her activities, although she perceived that she could do things to help herself. The depression scale indicated she was experiencing clinical depression, and had overall anxiety levels bordering on clinical significance. She had scores for separation anxiety, obsessive-compulsive features and generalised anxiety within the clinically significant range. Difficulties with depression and anxiety have been reported in studies investigating the emotional and behavioural consequences for children treated with brain tumours. Difficulties with peer relationships have also been reported but Heather did not report any difficulties for herself (Mulhern, Hancock, Fairclough & Kun 1992).

Follow-up assessment interview

The follow-up assessment with Heather and her mother revealed increasing concerns about Heather's inability to attend school although she clearly stated that that was what she wanted. Both were looking very unhappy and described their distress with the difficulty Heather was having making a full recovery.

Heather was suffering with recurrent and intermittent headaches, which she had experienced before the identification of a tumour and which started again as she became more mobile post surgery. She was seen for a neurological evaluation six months after surgery, and the results did not identify any neurological explanations for her headaches. However a study by Barr and colleagues (1999) investigating quality of life in brain tumour survivors, found that one third of 44 children reported chronic pain, confirming that this is a long term effect that has been reported by other children. At the time of the neurological

evaluation, it was noted that the varying level of intensity of the headaches were having an impact on Heather's school and home activities.

Heather was reporting headaches on most days typically occurring at the front and top of her head. Her pain worsened if required to concentrate on reading and writing and this made it difficult for her to undertake schoolwork. A recent MRI scan had indicated no evidence of tumour recurrence. Various medications had been tried with little impact on either frequency or level of pain from the headaches.

Both Heather and her mother were also concerned that she may have a visual field deficit. Heather reported that her vision went blurry if she watched television and she continued to find reading very difficult. She could not focus on any task that involved reading for any length of time.

Heather had had some psychological input with a view to focusing on using strategies to effectively manage her headaches but both she and her mother reported local referral to the CAMHS team was unhelpful.

At the time of the re-referral Heather was in year 7 at her local mainstream school. The transition from primary to secondary school has not been successful. Following her failure to cope with the initial transition, attempts were made to arrange a gradual introduction to the new school, but Heather had difficulty linking up with her friends and coping with the larger school. Increased noise levels and the greater number of pupils were also causing her distress. When she arrived at school no one seemed to know she was coming and she did not know where to find the classroom. The transition from junior to senior school meant a change in friends and as Heather was not in school from the beginning of the new term she did not establish a new friendship group. Her attendance at school was not

regular and very soon she stopped going. She continued receiving home tuition for two and half hours four mornings each week.

Since the initial assessment the family had undergone a period of severe upheaval and had been forced to move from the family home into a high rise flat. During this period Heather's brother was diagnosed with ADHD and given a statement of special educational needs. He also had significant reading difficulties and Heathers' mother was very distressed at the time it had taken for this to be identified and support to be put in place. As with her experience with Heather's illness, she had made repeated attempts over several years, to get the school to acknowledge her son's difficulties and get extra help for him. She was pleased with the support that had been agreed and with the school he would be attending.

Mother's expressed aim for coming to the appointment was twofold. She wanted further help for Heather to cope with her headaches, preferably medical help. Although both Heather and her mother reported she was making good progress and getting good academic support from the home tuition service, the education authority had recently informed them that this service would discontinue at the end of the academic year. Mother wanted support in her application for a Statement of Special Educational Needs to be put in place to help Heather return to school.

Hypotheses

Following this interview the original hypotheses were reviewed and new hypotheses developed to guide the approach to intervention for Heather and her family.

Now that time had passed since her diagnosis and treatment both Heather and her mother impressed as having low mood. Heather's depression and anxiety had been confirmed by the series of assessments carried out when she was in hospital.

It was hypothesised that Heather was experiencing difficulty returning to school because late effects from her treatment were impacting on her cognitive development and were also impacting on her ability to function in a busy school setting.

It was also hypothesised that Heather was suffering from feelings of hopelessness and depression and was feeling overwhelmed by the constant presence of her headaches.

As mother highlighted the difficulties Heather was having returning to school, a repeat psychological assessment was carried out to document any changes in cognitive functioning, which may impact on her learning. Literature has recognized that children two to three years past treatment with surgery only, show specific difficulties with learning and verbal memory (Levisohn et al 2000).

The literature has also reported long term effects on emotional behaviour including difficulty with self-esteem peer relationships, and low mood (Carpentieri et al 2003). Following the cognitive assessment a series of interviews were set up with Heather to provide advice and strategies to cope with the pain associated with headaches. These sessions were also used to consider self-esteem and mood.

A school visit was planned to meet with teachers, Heather and her mother, to consult about the effects of the tumour and about Heather's individual pattern of difficulties, with a view to guiding and advising on a school reintegration programme.

Second Neuropsychological Assessment

An assessment of cognitive functioning, numeracy and literacy was carried out using the Wechsler Intelligence Scale for Children-Third Edition^{UK} (Wechsler 1992), Wechsler Objective Reading Dimensions (Wechsler 1992, 1993) and the Wechsler Objective Numerical Dimensions. (Wechsler 1992, 1993)

Because memory, specifically verbal memory, has been documented as an area of difficulty

in children with cerebellar tumours and those who have had cerebellar mutism following surgery, the Children's Memory Scale (Cohen 1997) was used to provide a detailed assessment of Heather's memory. Executive functioning has also been documented as a potential area of difficulty (Spiegler et al., 2004) and as deficits in these skills could impact on Heather's ability to function both at home and in the school setting, mother completed the parent form of Behaviour Rating Inventory of Executive Functioning (BRIEF) (Gioia et al., 2000). Time constraints prevented a more detailed examination of this area and as Heather had not been attending school it was not appropriate to ask a teacher to complete the teacher's questionnaire.

The PEDsQL (Paediatric Quality of Life Inventory - Parent report (Varni 1998) and the PEDsQL (Paediatric Quality of Life Inventory-Teenagers report (Varni 1998) provided an assessment of emotional status and quality of life.

Results of Follow-up Neuropsychological Assessment

Heather presented as a cheerful, eager and willing young girl and approached the tasks with enthusiasm showing good attention and concentration throughout. There were times when she was experiencing discomfort due to a headache, specifically with tasks, which required reading and assimilating information. She was able to talk freely about her headaches and required short but frequent breaks.

Heather's profile of cognitive abilities indicated a decline in all areas of functioning since the baseline assessment three years previously. Full-scale scores had declined from the above average range to average range, a drop of 14 points. There was also a significant discrepancy (14 points) of the component parts (Verbal and Performance), which made up her Full Scale IQ score, in favour of her verbal ability, which was not present in the results

of the base-line assessment. (Details of scores from baseline and follow-up assessments are provided in the appendix.)

The pattern of declining scores was evident in Heather's index scores. Both her Verbal Comprehension and Perceptual Organisation index scores showed a decrease in comparison to her baseline assessment although both remained within the average range of functioning.

Heather's Freedom from Distractibility score of 104 indicated a significant change from her previous score (121). This index is made up from the Arithmetic and Digit Span subtests. She was having more difficulty with sustaining concentration while using short-term auditory memory for manipulating numbers.

Her Processing Speed index score of 78 was also significantly lower than her previous index score of 109, and her other index scores. The Processing Speed Index consists of visual-motor paper and pencil tasks, performed under timed conditions. Heather worked very slowly, although accurately on these tasks.

Attainments

Heather had made steady progress with all literacy tasks and mathematical reasoning, i.e. mathematical problems presented to her orally and visually. However, her score on the numerical operations, pencil-based exercises, has dropped since her previous assessment. Heather also mentioned that her headaches worsen when required to concentrate on writing.

Memory

Heather's general memory as measured by the Children's Memory Scale was within the high average range. Visual Memory was an area of strength. Her scores for both

immediate and delayed visual recall far exceeded her predicted level of functioning (as indicated by her WISC-III IQ score).

Heather found the recall of verbal material more difficult. Her ability to recall verbal information after its immediate presentation was within the average range, although less able than visual memory. Immediate verbal recall was commensurate with the level predicted from her WISC scores. However, after a delay, Heather was less able to recall verbal information. Her score of 85 on Delayed Verbal Memory was lower than what might be predicted from her Verbal IQ score, and less than her immediate verbal memory score. The results also indicated that Heather found verbal prompts relatively useful in facilitating her recall of verbal material after a delay, and her Learning index of 115 was within the high average range, which suggested that learning verbal and and visual based information over numerous trials had a positive effect on the amount she could recall.

Executive Skills

Heather's profile of executive capabilities measured by the BRIEF (parents form) indicated that she has difficulty regulating her own behaviour, including emotional control, shifting from one activity to another, and resisting impulsive activity. She was reported to have more difficulty with tasks, which required her to generate ideas and change activities. She was also reported to have difficulties with working memory. She was better able to organize tasks or activities, and monitor her own behaviour and achievements. Overall her composite score on executive functioning was within the clinical range indicating significant difficulties, which would be expected to impact on her daily functioning.

Pediatric Quality of life InventoryTM (PedsQLTM Varni 1998)

The Pediatric quality of life InventoryTM (Varni 1998) has been used extensively in the literature to assess quality of life of survivors of CNS tumours and their mothers (Eiser

2003). The literature has indicated differences in perception of the child's quality of life between mother and child. This has raised concerns about whose view is correct and how useful these measures are for reporting quality of life in children with chronic illness. However for the present case it was decided that in a clinical context any difference or concordance of views between Heather and her mother would be helpful in guiding future intervention.

The questionnaires (a parent and young person version) provide information on the child's present level of functioning in four domains: physical, emotional, social and school. Mother's report indicated continuing difficulties with physical activities including walking running and joining in sports or games. She described Heather as often feeling sad and unhappy, and angry or cross and having trouble sleeping at night. There were also indications from this report that Heather was having difficulty with social relationships, including problems getting on with her peers, difficulty keeping up with other teenagers during activities, and not being able to do things they can do. In school Heather had problems paying attention in class, keeping up with schoolwork, forgetting things, and having days off because of illness or needing to go to the doctor or hospital. Mother described difficulties in all areas of Heather's life.

Heather did not described any problems occurring as often as her mother did, but she added in headaches as being present almost all the time and highlighted the same areas on the physical scale as reported by her mother. She had trouble sleeping at night but she reported that she almost never felt sad or unhappy, or worried about what will happen to her in the future. She confirmed her mother's report that she often forgets things, had trouble keeping up with schoolwork, and taking days off school because she was not feeling well or had to see the doctor or visit hospital.

These scales provide an indication of the extent to which Heather's mother continued to have significant concerns about her daughter's quality of life and about the impact her illness was having on her daily functioning. Heather described similar concerns although not to the same degree as her mother. She was most troubled by her headaches. The response to items about her functioning within school, indicated difficulties, which may well result from impaired memory, slow processing sustained attention and difficulty with executive functioning such as emotional control and planning and regulating of her activity. This pattern of functional difficulty reflects the pattern of strengths and weakness reported from the neuropsychological assessment.

Post assessment interview

During a follow-up session to discuss the results from the neuropsychological assessment Heather confirmed that the headaches were a major concern. She accepted they would not stop but wanted help to be able to cope and return to school full-time. It became clear that the transition to secondary school, a larger and very busy environment, had proved too challenging and the level of support provided in school was not sufficient for Heather to manage the move. Functionally both she and her mother reported considerable difficulties arising from her verbal memory impairment. They were concerned that she would not be able to organise herself and find her way around school, or know which lesson to attend. This was an area of considerable concern for mother. Heather also remarked on the time she needed to develop her ideas and to complete written tasks. They both attributed the short time she was able to attend to written tasks to the significant effect of her visual difficulties.

It would appear therefore that despite good cognitive skills, the impact of her other difficulties were making it increasingly difficult for Heather to function at the same level as her peers, and to cope with the challenges of daily life.

Interventions and Outcome

The results of the second assessment have provided evidence that Heather has experienced considerable cognitive and functional deterioration in her ability to develop both her social independence and her ability to access learning alongside her peers and reintegrate back into school. This deterioration has been shown in the changing test results and in new symptoms of emotional and social dysfunction and isolation. Mother was also showing a high level of concern and it was clear could not help Heather reintegrate back into school if she was not satisfied with the type and level of support being offered.

It was hypothesized that when advising on appropriate intervention to help Heather it would be important to incorporate a range of social learning opportunities to help her address those activities of daily living that are restricting her ability to function independently, particularly in school. She rarely went out alone with friends and had become increasingly isolated from her friends in school, worrying about how to link up with them on those days when she did attend. However one of her expressed wishes for change was to be able to spend more time with her friends. The headaches provide a continuing focus of concern and served to restrict and limit Heather's attempts to reintegrate with school and her peers.

With this in mind recommendations to assist Heather in continuing to develop in all areas required not only direct focus on her headaches, the presenting problem, but also on helping her structure her day, develop social independence and facilitate her ability to access learning at all levels. A cognitive behavioural approach, which included a problem solving approach, was recommended for Heather. It was important that Heather had the opportunity to address her therapeutic needs, particularly around coping and adaptation to being diagnosed and treated for a life threatening illness. Her mother continued to talk

about the impact of this event and was understandably concerned for her daughter's future emotional and physical welfare. Cognitive behavioural therapeutic approaches with Heather would need to include the support and help from her mother.

Following the assessment a meeting was organized between Heather, her mother and appropriate school staff, including the Special Educational Needs Coordinator and the educational psychologist.

Recommendations

The following recommendations for ongoing support to assist Heather in her wish to overcome her difficulties and re-integrate fully back into school life were developed from the results of her neuropsychological assessment. Initially a high level of support, with a highly individualized programme both to aid learning and to help her cope with the more functional challenges of school life such as noise, moving from lesson to lesson, and planning her day etc was recommended. This would involve consideration of her problems with mobility, tiredness, and coping with her headaches. Heather would need a carefully graded programme, constantly reviewed and revised. Past experience had shown that she quickly lost confidence when feeling pressured and unable to cope.

More specific recommendations were developed based on relevant rehabilitation and remediation literature, drawn mainly from recommendations for children with acquired brain damage and developmental disorders. (Feeney & Ylvisaker, 2003; Rourke 1989; Ylvisaker & Feeney, 2003; Ylvisaker, Szekeres & Feeney, 1998). The recommendations were developed with the aim of increasing both adaptation to her difficulties and compensation, in an attempt to minimize the impact of her functional impairments. They also aimed to develop the awareness of teachers of the impact her pattern of difficulties would have on her ability to access learning. For example slow processing speed would be

expected to impede her rate of learning. Given her excellent memory for visually presented information it was important teachers were aware that Heather's learning would be optimised if the majority of teaching was conducted through this modality. However her specific difficulty with verbal memory particularly recalling verbal information after a delay would need to be considered in all approaches to new learning. Although she was aware that memory was a specific difficulty for her it was important to acknowledge that she would find it difficult to plan and evaluate strategies to overcome these difficulties and would need specific help to develop methods to support her recall of verbal information.

The results from the assessment of executive functioning indicated difficulties with emotional control, inhibition and shifting from one task to another. Each of these difficulties could be expected to impact on Heather's daily functioning and on her ability to adapt to changes in her daily routine. Given these difficulties in association with her memory difficulties it was important to ensure that her day was structured and predictable and remained consistent as far as possible. This was particularly important when planning her full reintegration into school. It was also important for the school to be aware that low and anxious mood would also impact on her ability to problem solve and cope with daily challenges.

It was strongly recommended that individual intervention to provide Heather with specific and ongoing help to cope with her headaches was made available. Her low mood would have an impact on her ability to adapt to changes in her life. Although a brief focused intervention was provided for Heather to develop strategies for coping with her headaches, these did not have a significant impact on her functioning and she decided not to continue with this intervention. As a result it was considered more appropriate to encourage this

intervention to be developed as part of the total package aimed at reintegrating her into school.

These recommendations were discussed with the school with a view to integrating them into academic support. As a result of discussions, Heather was referred to a locally based education support unit, where she could be provided with an adapted educational programme and also support with her emotional and social difficulties. Transport was provided to take her to the unit but not to take her home, another anxiety for mother. However Heather remained committed to the idea of returning full time to mainstream education and the family planned to pursue their request for a statement of educational needs with the aim of achieving full time individual support with an adapted educational programme within the mainstream setting.

Discussion

Heather's neuropsychological profile is in accord with the pattern of difficulties reported in the literature on children who have had surgery as the only form of treatment for a cerebellar tumour. Although she had cognitive skills within the average range she had specific difficulties with processing new information, verbal memory in association with slowed motor output and processing speed. There were also indications of poor attention and some compromise of visuo-spatial processing and regulation of affect (Carpentieri 2003; Levisohn 2000; Riva & Giorgi 2000). The above average cognitive skills indicated from the baseline assessment had not ensured that she would be able to return to school and function in her daily life at the same level she had achieved before her illness. The pattern of her difficulties was not apparent in the assessment carried out soon after treatment or in reports of her daily functioning at that time, but evolved over time. Detailed analysis of her present cognitive status together with other areas of functioning including

memory, executive functioning, emotional and physical status have gone some way to providing an explanation of her increasing inability to cope with daily life despite an expressed wish to reintegrate with her peers. The assessment was carried out under optimum conditions and provided an indication of the level Heather was able to achieve in quiet surroundings where she was encouraged and supported, and external noise or distractions were kept to a minimum. This was similar to the situation when she had her daily home tuition.

It is the multi-dimensional view that provides the information necessary to understand her difficulties and her ability to perform in different situations, and to guide intervention and rehabilitation strategies. Her pattern of cognitive difficulties in association with headaches and emotional difficulties were not immediately apparent when talking to Heather, and the school were unaware of their functional impact. Heather was able to present as a friendly positive girl and the school were frustrated in their efforts to help her reintegrate back into school, expressing the belief that she was not willing to cooperate. During the meeting with school staff it became apparent that they did not appreciate the subtlety of her difficulties nor that she would need a high level of very individual and structured support to re-establish an ordinary life. As a result there was a reluctance to support an assessment for a statement of educational needs. This situation was ongoing and complex and unlikely to be resolved without a high level of direct intervention on a long-term basis.

It is important that children who have been diagnosed and treated for brain tumours are monitored and reviewed not only post treatment but also some years later particularly at times of transition even where treatment has been minimal and the prognosis is good.

Although there are no reports in the literature of children who have been diagnosed and treated for brain tumours having difficulty with transition into secondary education, clinical

experience has confirmed that this is a time that is fraught with difficulty and requires careful proactive intervention to ensure that the necessary supports are in place.

This case demonstrates the view that a developmental approach to rehabilitation which reviews a child's functioning over time is required to fully assess the impact of diagnosis and treatment for a brain tumour, even in those children where treatment is considered minimal and the prognosis is good. As reported in the literature (Feeney & Ylvisaker 2003) Specific intervention directed at helping her cope with her headaches did not generalise and did not impact on her ability to cope with everyday life although they were considered the main obstacle to her return to school. Although Heather's reintegration into school is not yet resolved and the outcome of providing a more individualise support programme has not yet been assessed, the case also demonstrates the importance of including not only the family but also the educational services in plans to develop rehabilitation strategies.

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APPENDIX

Scores from the previous assessment are reported here to allow comparison of results.

1. Wechsler Intelligence Scale For Children - Third Edition (WISC-III ^{UK})

Verbal Scale Subtests	Scaled Scores		Performance Scale Subtests	Scaled Scores	
	2000	2003		2000	2003
Information	11	9	Picture Completion	14	15
Similarities	15	14	Coding	11	5
Arithmetic	16	11	Picture Arrangement	9	5
Vocabulary	9	12	Block Design	12	11
Comprehension	13	11	Object Assembly	12	13
(Digit Span)*	12	10	(Symbol Search)*	12	7

*Test scores not used to compute IQ score

	IQ Scores		Level of Functioning 2003
	2000	2003	
Verbal IQ	117	108	Average range
Performance IQ	112	96	Average range
Full Scale IQ	117	103	Average range

Index Scores

	Index Score		Level of Functioning 2003
	2000	2003	
Verbal Comprehension Index	111	108	Average range
Perceptual Organisation Index	111	104	Average range
Freedom from Distractibility Index	121	104	Average range
Processing Speed Index	109	78	Mild Learning Difficulties range

2. Wechsler Objective Reading Dimensions (WORD)

	Standard Score		Age Equivalents 2003
	2000	2003	
Basic Reading	87	97	11 years 9 months
Spelling	96	89	10 years 3 months
Reading Comprehension	100	98	12 years

3. Wechsler Objective Numerical Dimensions (WOND)

	Standard Score		Age equivalents 2003
	2000	2003	
Mathematics Reasoning	117	115	16 years 3 months
Numerical Operations	111	89	10 years 3 months

4. Children’s Memory Scales (CMS)

	Index Score	Percentile Rank	Functioning Level
Visual Memory Immediate	128*	97	Superior range
Visual Memory Delayed	122*	93	Superior range
Verbal Memory Immediate	109	73	Average range
Verbal Memory Delayed	85~	16	Low Average range
Verbal Memory Delayed Rec.	91	27	Average range
Attention and concentration	112	79	High Average range
Learning	115	84	High Average range
General Memory	117	87	High Average range

*Scored higher than what one might predict given H’s WISC 111 IQ
~Scored lower than what one might predict given H’s WISC 111 IQ

5. Behaviour Rating Inventory of Executive functioning (BRIEF)

	Index Score	Percentile Rank
Inhibit	70	94
Shift	81	99
Emotional Control	68	94
Behaviour Regulation Index	75	96
Initiate	79	99
Working Memory	65	91
Plan/Organise	43	33
Organisation of Materials	64	92
Monitor	52	65
Metacognition Index	60	85
Global Executive Composite	67	92

SECTION D

**COGNITIVE FUNCTIONING FOLLOWING DIAGNOSIS AND TREATMENT
FOR PAEDIATRIC BRAIN TUMOURS IN THE POSTERIOR FOSSA: REVIEW
OF THE LITERATURE AND SUGGESTIONS FOR REHABILITATION AND
FUTURE RESEARCH**

Improvements in treatment for children diagnosed with a brain tumour have led to better survival rates, with many more children surviving into adult life. For some tumours the survival rate is now 90% (Magnani 2001). With these improved rates, there is an increased awareness of significant long-term complications, both tumour and treatment related, on the child's continuing development. For children, the pattern of neurological and cognitive impairment might not be stable but is vulnerable to factors such as the age when diagnosed and treated, and continuing development. When assessing long-term effects, length of time since diagnosis, social environment and emergence of new skills are important considerations.

A recent report on outcome in a group of 21 children treated for brain tumours at Great Ormond Street Hospital has shown that 17 have neurological, psychological or cognitive difficulties, which are impacting on their day-to-day life (Gumley, Bruce, Phipps & Michalski 2004). A child with full scale IQ within the normal range can have specific difficulties and disabilities such as visual impairment and motor problems, which together with the child's individual neuropsychological profile will impact on their ability to access learning and to cope with the tasks of daily life. It is important to take a holistic view of the child, including the physical, cognitive and psychosocial factors, when considering the impact of tumour and treatment on the child's continuing development and to guide effective intervention and rehabilitation strategies. Longitudinal studies will provide more information on the natural progression of the effects of both the tumour and treatment and can also guide understanding of the prognosis for long-term cognitive and social affects on the child's ability to acquire and consolidate new knowledge.

It is therefore important to assess treatment and tumour sequelae over time, adjusting intervention and rehabilitation needs in line with the child's developmental progress and surrounding context.

Provision to facilitate both child and family adjustment to diagnosis and treatment and plan for their future needs, including social emotional adjustment and functional impact, needs to be a dynamic and proactive provision, which can plan for changing future needs.

Background

After leukaemia, intracranial tumours are the second most common neoplasm to occur during childhood. They account for 20% of all paediatric cancers (Tobias & Hayward 1989) and tumours of the cerebellum account for half of all brain tumours diagnosed in children. The posterior fossa is the most common region of the brain affected (Heideman, Packer, Albright, Freeman & Rorke 1997).

The estimated incidence of tumours in children under fifteen years is between 2.5 and 3.5 per 100,000. The rate of children diagnosed with a brain tumour, surviving 5 years past diagnosis when they are younger than fifteen years, has increased from around 35% in the 1960's to 60% and more in the present day, depending on tumour histology, age at diagnosis and country in which treatment is received (Magnani et al., 2001). The outcome for some tumours such as cerebellar astrocytoma has improved from 40% to more than 90%.

Brain tumours are classified by location and histology; the most common types are medulloblastoma, astrocytoma, ependymoma and brain stem glioma. They are graded according to degree of malignancy, from benign to malignant, which together with location and tumour type will guide treatment. However, the discrimination of difference between malignant and benign tumours is difficult, as both types of tumour can be destructive to neural tissue depending on rate of growth, invasiveness, and location. Tumours can compromise functioning through increased intracranial pressure, inducing seizures,

destroying brain tissue through invasion or replacement and by secreting hormones or altering endocrine patterns that affect a variety of body functions (Spoudeas 2002).

Guidelines for treatment are reviewed and provided by the United Kingdom Children's Cancer Study Group. Most tumours are treated first with surgery, which may include biopsy for a diagnosis, partial or complete resection of the tumour. This can be followed by whole brain or focused radiotherapy, and chemotherapy. For some children a 'watch and wait' policy may be adopted. Chemotherapy has become the treatment of choice for children under the age of three years. Variations in treatment will depend on the type and location of the tumour, and can extend over a number of years if the child has radiotherapy and chemotherapy. Routine MRI scanning for at least five years, (initially every three months, reducing to once in twelve months), is carried out for most children. Following the initial treatment period, monitoring may also include regular appointments with a range of medical specialties such as neurology, endocrinology, and ophthalmology.

The nature of treatment can create significant problems for children and their families, and although many children cope well, social cultural and family issues can influence outcome (Carpentieri et al., 1992; Mulhern et al., 1993). Cognitive dysfunction is common but the manifestations may vary widely and no distinct neuropsychological profiles have been described (Ris & Noll 1994; Mulhern et al., 1992). Research studies have extensively investigated and documented various factors such as age at treatment, extent of neurosurgery, presence of post-operative complications, development of hydrocephalus, adjuvant radiotherapy and tumour location that can adversely effect a child's future cognitive skills and educational performance. These factors can be associated with an increased burden of care for the family. There is a need to consider the quality of life for

these children and their families, and to increase our knowledge of long-term effects on psychological and psychosocial functioning.

However, there are problems with investigating treatment effects in this group of children because not only is this a rare condition, but there are a number of factors including type of tumour and location within the brain, which in themselves will influence treatment decisions and outcome. The literature is characterised by small numbers and in an effort to build up numbers, many studies include children whose progress has been influenced by a range of variables, which cannot all be controlled for in an investigation. Other studies report on very small numbers or wide age ranges and have not included control groups of typically developing children. However this is an important area of study as there are significant implications for the child's functional status and their ability to access education and develop positive peer relationships.

The present review will summarise the results of some of the early studies on treatment effects and cognitive functioning, but will concentrate on reports from the last few years, particularly in relation to cerebellar tumours, including medulloblastoma and astrocytoma. Survival rates for these tumours have improved so dramatically, it is important to assess whether changes in treatment have impacted on cognitive functioning and emotional and behavioural functioning. It is also important to report positive outcome but there are few studies in the literature, which have documented good outcome for this group of children.

Late effects have been defined as any chronic or late occurring physical or psychosocial outcome persisting or developing after diagnosis of the tumour (Anderson et al., 2001). Mulhern (1994) defined late effects as occurring after the successful treatment of the primary disease, differing from short-term effects such as nausea and vomiting associated with treatment modalities such as chemotherapy. Some investigators have defined five

years as the minimum period post treatment that an outcome can be defined as a late effect. However treatment effects can be evident during and immediately after treatment and will have an effect on the child's future progress from that point.

Many children have motor and sensory deficits and these will be considered first as they have implications for the child's long-term cognitive development and functioning. Implications of studies for future rehabilitation needs will also be considered.

Motor and Sensory Outcome

Children who survive brain tumours may suffer varying degrees of neurological impairment, including seizures, chronic pain, motor and sensory deficits. Differences in the incidence of neurological deficits have been reported and reflect the variations in tumour histology, malignancy, location, and treatment. Approximately 80% of children who have had brain tumours, experience some subsequent morbidity with more than one attribute affected in the great majority. They can have a complex pattern of neurological deficits, with associated behavioural and emotional difficulties. Children with optic pathway glioma and craniopharyngiomas have a high incidence of significant visual deficits associated with the tumour, including blindness, visual field deficits and perceptual deficits (Poretti et al., 2004). Visual impairment will have a significant effect on the child's future functioning and ability to access learning in the mainstream school. Children have also been reported to develop hemiparesis, cranial nerve deficits, and seizures following surgery. A study by Barr and colleagues (1999) which investigated quality of life in brain tumour survivors, found that one third of 44 children reported chronic pain. A recent study by Sønderkær et al. (2003) reported that although children who had surgery as the only form of treatment had a good chance of recovery, two-thirds would have neurological deficits. Among children with posterior fossa tumours, the frequency of children with partial to severe disability was

higher in girls than boys reflecting a higher frequency of ataxia and hemiparesis among girls. One third of the group had partial recovery of pre- and postoperative deficits and symptoms and one third continued to have severe neurological disabilities that required regular follow-up and high levels of physical and social support. Sønderkær et al. (2003) also found that whilst total tumour resection is an important prognostic factor for freedom from seizures they did not find any significant effects from age of diagnosis, although this has been reported in other studies (Jenkin, Danjoux & Greenberg 1998), duration of symptoms, or presence of hydrocephalus on the child's neurological outcome. Although, posterior fossa tumours are more likely to result in neurological deficits than tumours in other areas of the brain, neurological outcome is variable and it is important that a careful assessment is made for each individual child, when considering implications for rehabilitation and future learning needs.

Cognitive Outcome

Numerous variables, which may impact on cognitive outcome, have been documented in the literature and these will be considered in turn. Most studies have investigated the consequences of radiation therapy, but sequelae have also been noted in those children who have had only surgery, and those who have had chemotherapy.

➤ Surgery Only

The neuropsychological and neurobehavioral sequelae resulting from resection of cerebellar tumours in children is a developing area of study. Peri-operative factors such as neurological deficits, meningitis, shunt infection or the need for repeat surgery, have been shown to correlate significantly with IQ deficits in children following treatment for medulloblastoma tumours in the posterior fossa (Kao et al. 1994; Ater et al., 1996).

Studies, which have aimed to investigate the role of the cerebellum in cognitive and affective development in order to guide treatment and rehabilitation strategies, are

beginning to report a characteristic pattern of impairments. From the period immediately following surgery, children are showing impairments in executive functioning, planning and sequencing, visual spatial function, expressive language, verbal fluency, language sequencing, memory, including spatial and visual memory and regulation of affect and slowed processing speed (Levisohn, Cronin-Golomb & Schmahmann, 2000; Riva and Giorgi 2000; Karatekin, Lazareff and Asarnow 2000; Steinlin et al., 2003). The majority of these studies have investigated children with mean ages from seven years to ten years, and report that although the Full Scale IQ is within the normal range, children continue to have specific impairments at least two years post surgery. Visual spatial difficulties were characterized by impairments in planning and organisational aspects of tasks. Deficits in expressive language were characterized by brief responses, lack of elaboration, reluctance to engage in conversation, long response latencies and word finding difficulties. Riva and Giorgi (2000) reported more specifically, relating impairment to location. They indicated that children with right cerebellar tumours had disturbances of auditory sequential memory, and language processing, whereas left cerebellar tumours had deficits in spatial and visual sequential memory. Overall regardless of the laterality of the lesion the children in the study showed a slight worsening in naming and comprehension, and difficulties with processing of executive and time based attention tests.

The findings in the literature also support an association between extensive vermis damage and impaired regulation of affect, including irritability, impulsivity, disinhibition and lability of affect with poor attention and behaviour modulation (Levisohn, Cronin-Golomb, Schmahmann 2000; Riva & Giorgi 2000). Riva and Giorgi (2000) have examined this group of children more extensively and found that children with lesions in the vermis showed two profiles in the immediate post surgery period. One group of six children

showed post surgical mutism. Four of the children completely re-acquired normal speech over a period of six to twenty-four months but in two children the mutism evolved into speech disorders or language disturbances. Five children showed post-operative behavioural disturbances ranging from irritability to withdrawal from social contact and behaviour reminiscent of autism. The behavioural changes in four children gradually improved and returned to normal over a period of three to four weeks. Although the fifth child showed some improvement, and her behaviour settled down, she remained generally introverted. The evidence is therefore indicating that deficits are not a short-term response to the impact of surgery.

A further study by Carpentieri and colleagues describe the neuropsychological functioning after surgery in a group of 77 school-age children who were evaluated within three months of surgery for localised brain tumours, not all in the cerebellum (Carpentieri et al., 2003). Although the Full Scale IQ of the children was within the normal range, 45% showed a significant verbal/performance IQ discrepancy in favour of verbal IQ. They were showing more difficulty with non-verbal tasks.

The authors specifically documented the impact of processing and output speed, memory deficits and vulnerability to complexity, on functioning and concluded that as these children were assessed before receiving radiotherapy, other mechanisms such as the presence of the tumour, the impact of surgery or the surgical procedure could contribute to neuropsychological outcome. However the study group included a very wide age range and a range of tumours, so specific effects related to tumour or tumour site could not be documented.

The literature has indicated that children treated by surgery alone for tumours in the posterior fossa have full-scale IQ scores within the normal range. However, more specific

difficulties with language, attention, memory, processing speed and executive functioning, would be expected to impact on a child's ability to access learning and make positive academic progress. Difficulties with behaviour as reported by Riva and Giorgi (2000) and Levisohn et al. (2000) will also impact on the child's daily functioning and create additional stresses for family life.

➤ **Cognitive effects of Radiotherapy and Chemotherapy**

Review of published outcome studies of treatment effects on cognitive functioning in children treated for brain tumours, reveals cognitive decline in association with both radiotherapy and chemotherapy treatment (Hoppe-Hirsch et al., 1990; Jannoun & Boom 1990; Palmer et al., 2001). The rate of decline has been associated with younger age at time of treatment, time since treatment, female sex, and hydrocephalous, radiotherapy and radiation dose (Ris et al., 2004; Goldwein, Jones-Wallace & Boyett 2001; Armstrong et al., 2004). Children under the age of seven are reported to show a significant decline in IQ by one year following radiation (Radcliffe et al., 1994), with significant changes in scores three to four years from baseline assessment. Others have indicated that younger children show an immediate decline, with plateau at 6 years post diagnosis, and older children show some delay before a decline in skills (Palmer et al., 2003).

Although declines in IQ have been shown in children aged less than ten years who received cranial irradiation, young children who did not have radiation, and children over ten years have been shown to have stable IQ scores (Ater, Copeland & Moore 2004). Other studies have begun to look at the relationship between IQ and changes in the white matter of the brain following radiation (Palmer, Reddick, Glass, Mulhern 1999) and have begun to report on the presence of white matter lesions following radiotherapy, associated with neuro-cognitive decline (Fouladi et al., 2004).

Radiation effects have been associated with general intelligence, academic achievement, including numeracy and literacy, verbal knowledge and reasoning, and perceptual motor difficulties. Specific problems with executive function, attention, aggression, memory, slow processing and difficulty acquiring new learning, have also been reported. Methotrexate has been documented to cause neurological and neurobehavioural impairment and has been reported by Riva and colleagues (Riva 2002) to worsen cognitive deficits induced by chemotherapy and radiotherapy. Children who received methotrexate are also reported to do poorly in tests exploring executive functioning.

A review of the literature on the neuropsychological status of children treated for brain tumours by Mulhern, Hancock, Fairclough & Kun, (1992) identified 22 English language publications, representing 544 patients. They assessed the relationship between risk factors such as age at diagnosis, tumour location, and radiation therapy, time since completion of treatment, and subsequent intellectual development, academic achievement, psychosocial functioning and neuropsychological status. They also evaluated, where possible, other factors such as presence of seizures, and sensory and motor deficits. The results from the review confirmed the importance of radiation therapy volume, particularly whole brain radiation, and age at treatment on IQ. No effects were found for tumour location, but younger children treated with whole brain irradiation showed a 14-point deficit in IQ as compared with their older counterparts. Although there were no significant differences between older children receiving whole brain irradiation and those receiving local irradiation, both groups had IQ levels 12-14 points lower than those not irradiated. At the time of the review, only two studies had investigated the impact of visual, auditory and motor impairment. Although they reported no statistically reliable correlations between

these deficits and IQ scores, five of six children in one study (Mulhern & Kun 1985) with IQs below 80 had visual impairment.

Mulhern et al. (1992) carried out a multi-study analysis of IQ effects and found that children with posterior fossa tumours receiving cranial irradiation had lower IQ scores than children with no irradiation and also had lower IQ than children who had received focal irradiation. Children under the age of four years who had received cranial irradiation were at the highest risk for impaired cognitive functioning.

Other studies such as that by George et al. (2003) have also investigated cognitive sequelae in children with posterior fossa tumours. They reviewed memory and cognitive functioning in a group of 15 children diagnosed with medulloblastoma and cerebellar astrocytoma. The children were assessed an average of 3.5 years after treatment with radiotherapy. Their findings confirmed findings previously reported in the literature. The mean IQ scores and memory scores were significantly lower than those of the normative population. There were no significant differences between verbal and nonverbal IQ or verbal and visual memory in this group although the IQ scores of children diagnosed when less than six years were significantly lower than children diagnosed over six years.

Educational progress can provide a measure of functional outcome and although few studies have investigated academic achievement, those that have, reported significantly decreased arithmetic scores (Silverman et al., 1984) in children with brain tumours when compared with sibling controls.

Educational progress has also been used as an outcome measure by investigators such as Zucchinelli and Bouffet (2000) and Phipps (1996). Zucchinelli and Bouffet sent out questionnaires to 34 families of children treated during 1987-1993, who were under age twelve years at treatment and at least three years post diagnosis. In their group, twenty-six

children were reported to be experiencing learning difficulties, including slowness, memory and comprehension difficulties, which affected mathematics, reading, and spelling. There are therefore indications that reported cognitive deficits are having a significant effect on the children's ability to function well in the school situation.

Although there have been reports of decline relative to a normative population in IQ over time following radiation treatment, the literature has only recently begun to look at the nature of the decline in cognitive skills. The aim has been to consider whether the decline in scores represents an actual loss of skills and knowledge, or whether children are acquiring new knowledge at a slower rate than their same age peers. Palmer et al. (2000) investigated fifty-two children who had been treated with surgical resection, cranial radiation, with or without chemotherapy, for medulloblastoma in the posterior fossa. Each patient had had cognitive assessments at multiple times, with the most recent assessments 1.8 to 12.6 years following treatment. At the most recent assessment all outcome scores were at least 1sd below the expected population norms. The authors' analysed changes in the age adjusted standard scores for subtest on the WISC-III and WAIS, and revealed a significant decline over time in subtest scores. However when they examined raw subtest scores over time, they were found to significantly increase. Palmer et al. (2000) concluded that children treated for medulloblastoma were making progress in their learning but below expectations when compared with their peers. They did not differentiate between variations in the rate of new learning across different subtests.

Late effects from radiotherapy on Full Scale IQ have been well documented in the literature, but few studies report on those children who are unable to complete formal cognitive assessments, as a consequence of significant learning difficulties. Although specific aspects of cognitive functioning, such as memory or verbal fluency and attention

and executive function have also been comprehensively examined, there is little information on whether these skills show a decline over time along with full scale IQ scores or remain stable following the initial insult. Spiegler et al. (2004) reported significant declines in visual-motor integration, visual memory, verbal fluency and executive functioning following treatment with cranial radiation in a group of children, but no evident decline in verbal memory and receptive vocabulary. It is these more subtle aspects of changing cognitive skills that will impact on a child's adaptive functioning and perhaps contribute to the slow progress in learning reported by Palmer et al. (2000).

Emotional & Behavioural Effects

Long term neuropsychological deficits resulting from diagnosis and treatment effects of brain tumours could be expected to have consequences not only on academic functioning of children but also on adaptive functioning and on emotional and behavioural functioning both at home and at school.

In the studies that had investigated emotional and behavioural functioning, emotional adjustment has been reported to be a problem for a high number of children diagnosed with a brain tumour. Mulhern and colleagues have been investigating emotional and behavioural adjustment in a number of studies over a long period of time. At post-surgical evaluation they reported that 50% of children showed disturbances of behaviour not representative of premorbid functioning (Mulhern and Kun 1985). A review of the literature (Mulhern, Hancock, Fairclough & Kun 1992) concluded that children with brain tumours show at least some increase in social and emotional problems, with report of somatic complaints, social withdrawal and depression.

When comparing a group of children newly diagnosed with a brain tumour with a group of children newly diagnosed with other cancer, not involving the central nervous system, both

groups of children were shown to exhibit an abnormally high incidence of social and behavioural problems. Forty eight per cent of the brain tumour group had significantly greater functional impairment than the cancer control group, reporting moderate to severe functional limitations such as visual loss, hemi-paresis, and other problems with gross motor control. Tumour location, the child's functional status, and family situation such as single parent family and age of mother at birth of the child, were predictive of psychological status (Mulhern et al., 1993). Children with non-cerebral tumours and greatest functional deficits were found to be at the greatest risk of deficits in social competence whilst those children whose mothers were younger at the time of the child's birth were at greatest risk for behaviour problems.

However these investigations report the child's emotional and behavioural status in the immediate period after diagnosis and it is not surprising that there is evidence of emotional or behavioural disturbance in the presence of recently diagnosed life threatening illness. Of equal concern, particularly to the child's continuing functioning and cognitive development, is the child's emotional and behavioural status in the long term following diagnosis and treatment.

There is a body of evidence reporting on emotional and behavioural difficulties, in addition to documented cognitive difficulties, confirming that outcomes following treatment can be complex and long-term.

Hoppe-Hirsch (1990) reported that children with medulloblastoma continued to have psychological problems many years after treatment. Nearly half of the group of 55 children had problems with instability, slowness, anxiety, inhibition and negative attitude, up to five years after treatment and these problems were present in 78% of thirteen children reviewed

ten years after treatment. However they did not specify age at diagnosis, nor methods used to assess psychological status.

The surviving group of forty children from the Mulhern et al (1993) study were followed up and compared with a group of non-CNS cancer children (N = 40) when they were two to three years out from initial diagnosis (Carpentieri et al., 1993). Although both groups had higher scores on the Behaviour problems scale of the Child Behaviour Checklist (Achenbach 1991) relative to non-clinical populations, the brain tumour group did not have increased scores, and evaluation suggested the brain tumour group showed a significant decrease in emotional disturbance since diagnosis.

Fuemmeler, Elkin and Mullins (2002) carried out a review of 31 studies investigating psychological adjustment and quality of life in children, who survived brain tumours. They looked at general psychological adjustment, internalising behaviour problems, externalising behaviour problems, social competence correlates and predictors of adjustment and quality of life. The children appeared to be at risk for compromised social competence and long-term quality of life, but reports on psychological adjustment varied widely. Glaser, Rashid, Walker & Walker (1997) used the HUI (Feeny, Furlong, Boyle & Torrance 1995) to compare teacher's reports of children with brain tumours and their siblings to matched healthy peers and siblings. Teachers perceived children with brain tumours as having significantly greater impairments in emotion, worry, pain, and poorer self-esteem, than matched healthy controls. The children also had more mobility problems and were less likely to participate in formal sports. They did not have worse behaviour.

The psychosocial functioning and behaviour as described by child, parent and teacher was described in a cohort of 32 adolescents aged 12 – 18 years who were evaluated between one to five years post treatment for brain tumour (Carpentieri et al., 2003). They used the

self-report, parent report and teacher-report questionnaires of the Behavioural Assessment System for Children (BASC). Parents reported concerns about attention problems, and leadership; teachers had concerns about learning problems and both teachers and parents indicated somatization behaviours.

Fuemmeler et al. (2002) mentioned that those studies, which used standardized measures, found little or no evidence of internalising difficulties, whereas those employing interviews found greater evidence of these problems. As standardized measures are developed for normal populations and aim to distinguish clinical and psychiatric levels of difficulties they may not reflect the subtle pattern of difficulties found both in paediatric populations and particularly in children who have been treated for brain tumours. A study by Braun, Daigneault and Montour-Proulx (2002) investigated psychosocial functioning in children with focal unilateral cortical lesions using the eight scales of the Child Behaviour Checklist Parent Form (CBLC). The internalising, externalising scores did not dissociate as a function of lesion side, and they reported more internalising than externalising symptoms. Only the attention problems subscale yielded a significant difference in function related to lesion side, with children with left lesions presenting more difficulties. Braun et al. (2002) concluded that the CBCL reflects few of the characteristics of focal lesions in children and suggest that most of the elevations in CBCL scores seen in children with brain lesions could derive from the parents and child's emotional response to the medical situation and is not therefore measuring brain behaviour links.

Few studies relate emotional and behavioural difficulties to the child's cognitive functioning. Recently, Holmquist and Scott (2002) investigated emotional and behavioural outcome in children treated for brain tumours and treated with cranial irradiation, or chemotherapy. They investigated the association between time, age, and cognitive variables

and externalising / internalising behaviour as measured by the CBLC (Achenbach 1991) and the Conner's parent rating scale (Conners 1997). They indicated that children receiving certain types of chemotherapy were at risk for late onset of emotional and behavioural problems, including social withdrawal, somatic complaints, anxious and depressive symptoms, social problems and difficulties with attention. Children with deficits in cognitive functioning and verbal fluency were more likely to exhibit hyperactive behaviour than those without intellectual and verbal fluency difficulties.

Fuemmeler et al. (2002) found few studies, which investigated the presence of externalising behaviours such as hyperactivity, impulsivity, defiance or disruptive behaviour. A recent paper by Nakaji, Singel and Alksne (2003) reported two children who had right temporal lobe tumours together with significant behavioural abnormalities, including aggression, and anti-social behaviour. After surgical resection of the tumour both children's aggressive behaviour decreased markedly. Nakaji et al. (2003) recommend that when a brain tumour is identified in a child with behavioural abnormalities the possibility of a link between the two should be considered. Careful observation and assessment of a child's behaviour can therefore have direct diagnostic and treatment implications.

Steinlin et al. (2003) also reported that 33% of 24 children who were treated by surgery only, had behavioural difficulties such as attention deficit problems, mutism, uncontrolled temper tantrums and phobia, in addition to deficits in specific areas of neuropsychological functioning. A retrospective review of data from semi structured clinical interviews with 34 'surgery only' children, who had been treated for brain tumours and were two weeks to 5 years off treatment, was carried out by Meyer and Kieran (2002). The results indicated this group of children might be experiencing significant short and long-term mood, behaviour and adjustment problems when compared with the typical child population.

However the range of time off treatment is so wide in this study it does not distinguish between those children who are within the immediate period after diagnosis and those who have had a longer period of adjustment. Fuemmeler et al. (2002) also considered the evidence for difficulties with social competency, including adaptive behaviour, social skills, and peer relations, reported in children with brain tumours. Neurological deficits, including visual and auditory deficits, and slow processing of new information, may impact on the child's ability to interpret and respond to social cues. Age appropriate peer relationships are an important part of a child's adjustment to chronic illness and can have a moderating effect. Fuemmeler et al. (2002) looked at nine studies dating from 1976, which investigated different aspects of social adjustment in children with brain tumours. Children with brain tumours had significant deficits in social competency and were rated lower on these skills by their parents than children with non-CNS cancers. Few studies, have attempted to link cognitive outcomes, for example IQ scores, with emotional and behavioural outcomes or problems with school adjustment and learning.

Literature has also indicated that any child's progress and educational potential will be influenced by factors such as genetic endowment (Eysenck & Kamin 1981) home environment including parental attitudes to education (Haynes & Comer 1990), family stability or instability, socio-economic and cultural factors (Chevannes 1989; Dalal 1991; Haynes & Comer 1990) and the individual temperament of the child (Fontana 1983). Parental attitude to illness including family styles of coping both with sudden unexpected deterioration and change in their child's situation and possible subsequent chronic illness also requires consideration. The parent's ability to adapt to events and ongoing pressures affects the child and other family members in different ways. Interaction between family members has been shown to have an impact on the child's self image, behaviour and

developmental progress. Coping strategies adopted by individual parents are variable and include how they perceive threatening experiences or stressful events, how they manage their emotions and how they attempt to solve problems (Eiser 1998; Grootenhuis & Last 1997; Sawyer, Streiner, Antoniou, Toogood & Rice 1998).

These family factors were considered, in a study investigating the prediction of cognitive and behavioural outcomes in children with heterogeneous brain tumours (Carlson-Green, Morris, Krawiecki 1995). Family and demographic variables were found to be the best predictors of children's behaviour problems and adaptive behaviour. Parents reported a slightly elevated rate for behaviour problems. Family stress levels were positively associated with the number of child behaviour problems reported. Children in two parent homes from families that had experienced fewer life events had significantly fewer behaviour problems. A combination of family and illness variables was the best predictor of intellectual functioning. These results provide some support for the inclusion of contextual factors such as family stress, maternal coping, number of parents in the home and family measures in studies of how disease factors affect outcome in children with brain tumours.

Demographic, medical, cognitive and psychosocial measures at short-term outcome (one to two years post diagnosis) were used to predict long-term social and behavioural outcome, in 40 children with brain tumours (Kullgren, Morris, Morris & Krawiecki 2003). Kullgren and colleagues aimed to predict social and behavioural outcome three to four years post diagnosis. Below average social competence and average behavioural ratings were reported at follow-up. Multiple medical treatments and lower social competence scores at initial assessment were predictive of long-term difficulties concerning social competence. A series of qualitative interviews with parents of eight children who had been diagnosed with medulloblastoma, (Vance & Eiser 2004) highlighted the complexity of

problems experienced by the children and their families, including inadequate educational support and difficulties with peer interaction. The children's physical problems were also reported to have an impact on their psychological functioning.

Although investigations inform about the likelihood of children with brain tumours suffering social emotional and behavioural problems, there have been few attempts in the literature to link the patterns of these difficulties to the site of the tumour, endocrine function, and neurological deficits or to treatment experienced by the children. These brain behaviour links could be expected to inform more directly on intervention techniques and provide opportunity for a more proactive approach to advice and information for the children and their families. Social competence, social interaction and academic skills become more of a focus of concern for parents with increasing time since treatment, with resulting anxiety about the child's future adaptive functioning and potential for independent living.

Studies have indicated that children with chronic illness and serious illness are at greater risk for psychological difficulties, but few studies, have investigated the implications of the additional risk of compromised cognitive and neurological functioning on a child's emotional and behavioural well being.

Intervention and Rehabilitation

Many factors are involved in predicting a child's recovery and quality of life following treatment for brain tumour. Although studies have indicated specific aspects of impaired cognitive functioning, few studies have addressed the implications the impact of changes in cognitive skills may have on the child's future adaptive functioning, nor the implications for rehabilitation. There are also few, if any studies, which explore the extent to which poor outcome is related to quality of, or access to, rehabilitation following treatment.

Assessment of functional independence in the children is complicated by the inherent difficulty in distinguishing between the direct damage caused by the brain tumour itself, effects caused by irradiation, and other treatment modes and the effects of increased intracranial pressure. Strategies for rehabilitation will need to be adapted to take account of comorbid difficulties such as attentional and sensory problems or physical impairments. It is also important to acknowledge that a high level of involvement and cooperation is required from families and from educational services if rehabilitation strategies are to be effective.

Studies (Varni, Blount, & Quiggins 1998; Katz & Varni 1993) have indicated non-familial social support significantly predicts the adjustment of children with newly diagnosed with cancer and may be enhanced through interpersonal social skills training. School reintegration programmes are also highly rated by parents as being perceived as helpful both for parents and the child (Katz, Rubenstein, Hubert & Blew 1988; Katz, Varni, Rubenstein, Blew & Hubert 1992; Frappaz et al., 2001). Recently a study by Sahler and colleagues (Sahler et al., 2002) indicated that the introduction of problem solving therapy had an impact on improving constructive problem solving in mothers of children recently diagnosed with cancer including those diagnosed with brain tumours.

However, few studies have assessed the effects of providing an intensive support programme on specific cognitive long-term effects of brain tumours and their subsequent treatment on children. Riva & Pantaleoni (1989) and others have demonstrated that focused and selective attention deficits in association with memory deficits can have a significant impact on the child's ability to recall information and can affect all areas of learning. Riva found that strong motivation activation improves the attention of children with posterior fossa tumours. Other studies have revealed that major recovery of lost skills does not occur without specific neuropsychological or social training (Emanuelson von

Wendt, Lundalv, Larsson 1998), and there is a need for ongoing rehabilitation. Intensive and immediate physical rehabilitation has also been demonstrated to be essential for laying down the foundations of cognitive rehabilitation (Philip et al., 1994; Puliyaodil et al., 1994). Strategies to overcome specific neuropsychological difficulties, which may be having an impact on the child's behaviour and educational attainments, have mainly been confined to the literature on children with traumatic brain injury, and there have been single case reports of interventions to address verbal memory deficits. Butler and colleagues (2002) described an outpatient based rehabilitation programme for children treated for cancer aimed at improving attentional and other neuropsychological deficits. Following intervention, the children showed statistically significant improvement on all measures of attention but there were no statistically significant changes on arithmetic achievement tests, an area of specific difficulty reported in children following treatment for brain tumours.

There has been a developing interest in remediation of memory problems, although this has been mainly focused on the adult population and there is a lack of child-specific strategies for memory rehabilitation. Approaches can include environmental adaptation, new technology, new learning together with emotional, behavioural and cognitive interventions. There are indications that remediation may have an impact on memory and reading comprehension deficits (Franzen, Roberts, Schmits Verduyn & Manshadi 1996). More recently researchers have begun to investigate the potential for interventions to bring about changes in executive functioning. Although much of this work has so far been confined to the adult population there have been indications that improvements may be possible particularly when focusing on developing strategies or external aids to help compensate for difficulties (Evans 2003). An intervention which included cognitive behavioural and an executive function routine was reported to have positive long-term

effects on reducing challenging behaviour in two children following traumatic brain injury (Ylvisaker and Feeney 1996) Other reports on interventions with children following traumatic brain injury suggest that cognitive rehabilitation and support directed at impairments in executive functioning, needs to be integrated into the child's daily life, both at home and school. New skills and coping strategies appear not to generalize unless training is related to functional skills (Ylvisaker, Szekeres & Feeney 1998; Ylvisaker & Feeney 2003). There are no reported studies documenting the use of such interventions in children with brain tumours. A further study has reported on the impact of introducing strategies to overcome slow processing speed in an adult male, with positive results (Wilson 2002). Slow processing speed has been reported in the literature on cognitive effect of brain tumours but there have been no studies reporting on the impact of direct intervention on this area of functioning.

Although there is extensive information on intervention and rehabilitation strategies for both adults and children, there is very little information on the impact of individual support and rehabilitation programmes for children with cognitive emotional and behavioural deficits resulting from brain tumour diagnosis and treatment. However, more studies are now reporting on interventions specifically aimed at providing social skills training for children with brain tumours (Die-Trill et al., 1996). A recent study by Barakat et al. (2003) evaluated the effectiveness of a social skills training group intervention to improve social skills and social functioning in children treated for brain tumours and to assess the impact of cognitive functioning on the effectiveness of the intervention. They found social skills and social functioning improved and that higher verbal and non-verbal functioning was associated with a higher response to the training groups. Another study by Barakat and colleagues (2003) found that a one-day family group intervention, which combined

cognitive behavioural and family therapy approaches, showed a decrease in symptoms of post-traumatic stress and anxiety.

There is therefore the potential for direct intervention to have an impact on the child's ability to function both socially and in an educational setting. This would require a multidisciplinary approach and close links between both school and parents and other professionals involved in the care of the child.

Discussion

A summary of the literature investigating the long-term effects of brain tumours has indicated the potential for significant cognitive deficits, which can be evident immediately post surgery, before any adjuvant treatment, and can also emerge over time. The literature has indicated that even those children who have undergone surgery only, will have a high chance of long-term neurological deficits. They often have residual physical disabilities (visual, hearing disability, hemi-paresis etc.) in association with other conditions related to their illness, such as tiredness or headaches. Neurological and sensory deficits will impact on a child's cognitive functioning. The children can also be subjected to a range of treatments and ongoing relationship with the medical system, which extends over several years with consequent regular absences from school. The impact of a child's neurological and sensory deficits on their ability to access learning, including time missed from school, will need to be considered when assessing treatment effects on long term function of these children. Literature has also suggested that low self-esteem and poor body image can influence school attendance and performance in normally developing children. These factors and their impact on both the child's and family coping strategies need further investigation. Clinical experience has indicated some children have difficulty returning to school when coping with changes in appearance and changes in motor function.

Overall the picture emerges of children with cognitive skills within the average to low average range. Throughout the literature there is a dependency on the use of Full Scale IQ scores or specific neuropsychological tests measuring specific aspects of cognitive functioning such as memory and attention, to assess treatment effects. The Full Scale IQ would suggest that these children are able to cope within the education system and with daily living skills, equally well as their peers who have not been treated for brain tumours. However when considering the other neurological, motor and sensory deficits, and specific neuropsychological deficits, which are reported in a high number of children, the full-scale score masks the particular difficulties children may face and the impact this will have on their daily lives. Clinically each child presents with a unique pattern of strengths and difficulties, which can be subtle and not immediately apparent to the classroom teacher or to their families. The difficulties can have significant effects on both the level of care children require and the amount of extra support they will need in an educational setting, to enable them to continue to learn new skills and develop appropriate strategies to function effectively into adult life. Although the main IQ assessments used are the Wechsler scales, there are few attempts to investigate the profile of strengths and difficulties shown by the children. Few investigations have included a complete WISC 111 for each child and there have been few reports in the literature, which used the index scores, to provide a more detailed analysis of patterns of cognitive functioning. Full Scale IQ is a global score, which evens out the difficulties and does not provide information on the effect other impairments such as visual or hearing impairments, memory deficits, executive functioning deficits, or slow processing, will have on the child's functional abilities. Whilst it is important to describe the cognitive functioning of children with different tumours in different sites, an assessment of late effects needs to investigate the impact of treatment on their daily lives

and to take a developmental perspective. Measures of quality of life go some way to address this problem but do not link to the functional reality of IQ scores.

Emotional and behavioural difficulties, which have developed in response to site of the tumour, will also affect the way the child is functioning in school and at home. However the literature has indicated that empirical measures of behaviour and social interaction do not reflect the picture provided by more qualitative measures or by considering the effect cognitive and language difficulties may have on emotional and behavioural status. For example impaired expressive language skills can restrict the child's ability to formulate and communicate ideas and feelings and thus interfere with the ability to cope with a range of social situations. These are children whose cognitive functioning is described within the average/low average range but who have a number of hidden disabilities which have significant consequences on their ability to function independently and learn in the same way as peers with the same overall level of ability. In assessing long-term treatment affects it is essential to consider the whole child and how deficits and strengths, including intellectual, emotional, behavioural and physical functioning, will interact to influence functional outcome for the child and their family. A multi-dimensional approach to the long term implications of diagnosis and treatment from brain tumours where factors, which may affect outcome, are assessed at different times in a child development may provide the best way of assessing the importance of different risk factors for the child's ongoing development.

Future Research

There are difficulties associated with studying the long-term effects in this group of children, particularly due to small numbers and the range of variables, which may influence outcome. For example, in an attempt to increase numbers, many early studies have tended to included a wide age range in their groups and children with a number of different tumours Both age

at diagnosis and age when a child is included in an investigation together with site and type of tumour will impact on functioning in a variety of ways. However this review has indicated that collectively the literature is providing a consistent picture of outcome.

Equally there are difficulties investigating outcomes of rehabilitation and intervention because of the individual pattern of difficulties seen in each child. Reports tend to be case descriptions drawn from the literature on children with acquired brain damage.

There is however consensus that the majority of children, who have been treated for brain tumours, either by surgery only or by the full gamut of treatment strategies available, will have long term affects on their emotional, behavioural and cognitive functioning. Further research is required to assess how responsive these difficulties are to individual rehabilitation programmes. It is also important to assess how proactive intervention can have a remedial effect for example on potential difficulties such as memory or slow processing speed which will affect the child's acquisition of new knowledge. Emotional and behavioural adjustment of children and their families is also recognised as an area that will impact on the child's future development. Although there have been studies reporting positive outcome for both children with brain tumours and their families, further investigation is needed into both the timing and effectiveness of intervention in all areas to guide future development of support services for this increasing population. Provision to facilitate both child and family adjustment to diagnosis and treatment and plan for their future needs, including social emotional adjustment and functional impact will need to be dynamic and proactive, planning for changing future needs.

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