The Executive Function and Theory of Mind Performance of Boys with Fragile X Syndrome

Volume One

A portfolio of academic, clinical and research work.

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Submitted in respect of the degree Psych.D. Clinical Psychology,

Course Code 23

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Introduction to the Portfolio

This Portfolio is a selection of work completed over three years, to satisfy the requirements for the degree Psych.D. Clinical Psychology. It is presented in two volumes. The first volume is divided into three chapters comprising academic, clinical and research work. All work is presented in chronological order. The pieces of work selected cover a range of client groups seen and perspectives and models used in clinical psychology.

The academic chapter is presented first. It contains five essays, a selection of those completed during the course. The chapter includes essays from the core areas of learning disabilities, child and adolescent and older adults, an essay on a neuropsychological topic and finally a essay from an area of specialist interest.

The clinical chapter contains summaries of the six clinical placements completed during the course, and abstracts of five selected case reports which are presented in full and in confidence in the second volume of this Portfolio.

The final chapter contains research completed during each year of the course. It contains small and large scale empirical pieces of work completed during the course, as well as a literature review and a group evaluation completed whilst on placement.

All pages in this volume (with the exception of some appendices) are consecutively numbered. To aid orientation, the title of each piece of work is given in the top right hand corner of each page.

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Summary of Academic Chapter

The academic chapter contains five essays, covering a number of topics of core and specialist interest in clinical psychology. The first is a critical discussion of the differences between Gentle Teaching and applied behavioural analysis as interventions used with people with learning disabilities whose behaviour is a challenge to services.

The second essay is a discussion of the applicability of cognitive behavioural techniques to child and adolescent psychological problems. This includes an overview of the nature of child and adolescent problems and the rationale of applying different cognitive-behavioural techniques to them. It concludes with a review of recent studies which examine the efficacy of cognitive-behavioural interventions with this client group.

The older adults essay discusses issues in providing group therapies for cognitively intact and impaired older adults. Process variables, which are assumed to be age-independent, are discussed prior to a consideration of those areas of groupwork which are unique to older adults.

The neuropsychology essay concerns the potential for clinical neuropsychology to contribute to the assessment of fragile X syndrome, and discusses a number of domains to which neuropsychology could contribute.

The final essay concerns the impact on children of having a sibling with learning disabilities, and discusses common methodological flaws in this area of research prior to critically reviewing recent studies of relationships between children and siblings with learning disabilities.

People with Learning Disabilities Essay

Discuss the use of a gentle teaching approach with people with learning disabilities who show challenging behaviours. Are any of the techniques used in gentle teaching common to those used in applied behaviour analysis?

Assessment A9

..... a culture of death..... is emotionally destructive, but slowly eroding the spirit rather than quickly consuming it. Behaviour modification is a perfect technology for a culture of death since it is based on control..... it asks for no substantive change from us, only technical competency.

McGee & Menolascino (1991)

.....Gentle Teaching is consistent with mainstream behavioural analysis.

Mudford (1985)

.....procedures such as Gentle Teaching may primarily involve the use of negative reinforcement to increase compliance.....For some, Gentle Teaching may not be very gentle at all....

Emerson (1992.)

Challenging behaviour is one of the major service issues for clinicians working with people with learning disabilities. Two competing approaches to tackling this problem are applied behavioural analysis and Gentle Teaching, which John McGee is credited with introducing. Over the past ten years, discussion between the proponents of both sides has been acrimonious, with Gentle Teaching supporters characterising behavioural analysis as being part of a “culture of death” (McGee & Menolascino, 1991). Gentle Teaching advocates have been dismissed as unscientific (Bailey, 1992) or offering nothing not already available through behavioural analysis (Mudford, 1985). Gentle Teaching has even been presented as a controlling and aversive procedure - the very antithesis of McGee’s stated aims.

The philosophy of Gentle Teaching

One of the main criticisms of Gentle Teaching, voiced by many researchers, is that it is extremely difficult to define exactly what is meant by Gentle Teaching, what its central

assumptions are and how one might employ a Gentle Teaching approach in practice. Whilst Jones & McCaughey (1992) view the changes in description of Gentle Teaching as being characteristic of a new and evolving philosophy, Bailey (1992) accuses them of being apologists for “dramatic flip flops in philosophy” (p881). A recent formulation of Gentle Teaching states:

Gentle Teaching is:

- The first step in creating feelings of companionship
- A set of strategies that encourages unconditional valuing and human engagement
- An approach that calls for mutual transformation
- An ongoing way of interacting
- A prelude to a psychology of interdependence

p8, McGee & Menolascino (1991)

Here, Gentle Teaching is described by a mixture of processes (“an ongoing way of interacting”), philosophy, and a means to an end. It draws heavily on abstract concepts (“mutual transformation”, “psychology of interdependence”). Thus, the first point of note is that it is not operationally defined. The reader is left unenlightened on ways of employing Gentle Teaching in practice. Therefore, it is helpful to consider Gentle Teaching in terms of its core beliefs and assumptions. Jones & McCaughey (1992) identify these as bonding, communication and valuing.

Bonding:

McGee & Menolascino (1992) state that:

“ A psychology of interdependence...is based on the belief that all of us long to be companions in this life, and this feeling of being with others and a sense of belonging resides in all of us”.

Thus Gentle Teaching, being a prelude to a psychology of interdependence, encapsulates the belief that companionship and belonging are central to human nature. McGee & Menolascino go on to consider the bonding and attachment that occurs between mother and infant as being where we learn the value and essential goodness of human interaction. Their idea, then, is that those marginalised by society in general, including people with learning disabilities, have not learned the value of human interaction and reciprocity. This is seen as an important cause of the development of challenging behaviour.

This view has much in common with psychodynamic interpretations of the behavioural problems commonly exhibited by people with learning disabilities. Gaedt (1995) describes how the early interactions of people with learning disabilities may be hindered by sensory or cognitive impairments, thus preventing the attainment of developmental milestones. He considers that these may manifest later in life as a deficit of mature ego functions and ego structures.

As a deficit in bonding is seen as central to the aetiology of challenging behaviour, one of the aims of Gentle Teaching is rectify this deficit. The process of teaching bonding, according to Gentle Teaching, requires that mutual change occurs in the caregiver and client. It is not sufficient for a carer to change only their overt behaviour, Gentle Teaching requires "our values...to be constantly questioned and deepened" (McGee & Menolascino, 1991). Thus, Bailey's (1992) claim that all behavioural interventions require change on the part of the carer does not seem to require the same magnitude of change as McGee & Menolascino advocate.

The notion of bonding has been heavily criticised by behavioural analysts (Bailey, 1992; Cuvo, 1992). Their objections centre around what is meant by the terms as they are used throughout the Gentle Teaching literature. If the terms are meant to be descriptive rather than explanatory, then they may offer a useful insight into the development of behavioural problems in this client group. However, if bonding is

intended to be an experimental variable in a programme using Gentle Teaching then the definitions given are too soft to be useful. Observable variables which McGee uses to characterise bonding, such as praise, cuddles and other forms of approval, can be re-labelled as social reinforcement by behaviour analysts (Mudford, 1985).

Communication:

Gentle Teaching draws heavily on the communication metaphor to explain challenging behaviour. In the earliest description of Gentle Teaching, challenging behaviour was viewed as a communicative message in response to a world perceived as meaningless and bewildering (Casey, McGee, Stark & Menolascino, 1985). Some behaviourists share the assumption that all behaviour has message value (LaVigna & Donnellan, 1986), although would not share the same explanation for challenging behaviour.

The general assumption that all behaviour is an attempt to communicate has been criticised (Bailey, 1992; Emerson, 1992). Relabelling all behaviour as communicative has limited theoretical value if it does not distinguish behaviours which have “functional intent” i.e where the person intends to communicate something, from those where meaning is inferred by an observer without it being intended by the perpetrator. A further problem for Gentle Teaching is that behaviours are only allowed to carry one message according to their philosophy, which can only be remedied by teaching bonding. Functional analysis of behaviour indicate that a given behaviour can have a number of functions or outcomes for an individual including self-stimulation, tangible consequences, social attention or escape (Durand & Crimmins, 1988). It is the observation that behaviour can sometimes be maintained by escape from social interaction that implies that Gentle Teaching methods may be aversive for some clients (Emerson, 1992; Jones, 1994).

Valuing:

The final core belief of Gentle Teaching is that of valuing. McGee & Menolascino (1991) describe it as “a central task of caregiving” (p 32) and emphasise that it is a reciprocal process, where the caregiver has to seek valuing from the client. Again, valuing is not operationally defined, but McGee & Menolascino offer some insight of what valuing is in practice. It is:

- Given, not earned, during good moments and bad
- Centred on dialogue, that is, bringing the caregiver’s life into the world of the marginalised person’s through words, gestures, and physical interactions
- Involved in uplifting the person’s spirit and sharing

In behavioural terms, then, valuing is unconditional, and not contingent on behaviour from the client. It emphasises again the interactive nature of the relationship between client and carer, and the degree of involvement and dedication required of the carer. There is widespread agreement that staff valuing clients is a desirable and necessary part of services for people with learning disabilities. Bailey (1992) questions the legitimacy of considering valuing as unique to Gentle Teaching advocates, and considers their view of a person having intrinsic value just by being a human being as having no significance as it is so widely accepted.

Although it is accepted that people with learning disabilities are valued members of society it is not explicitly stated in the behavioural literature. This is a point Bailey (1992) goes on to concede stating:

“If there is any value to Gentle Teaching , it should be in prodding behaviour analysts to make their value system clear”

p882 Bailey (1992).

The explicit statement of a person's value is unique to Gentle Teaching, as is its assertion that valuing should be unconditional.

The main difference between Gentle Teaching and an applied behavioural analysis approach is the lack of operational definition of the central terms and constructs used by Gentle Teaching advocates to describe their approach. This is an anathema to behavioural scientists whose methods rely on the measurement and observation of behavioural signs, and has been interpreted as "Gentle Teaching's total disregard for science and the scientific method" (Bailey, 1992). Furthermore, Gentle Teaching's central notion of "bonding" has much in common with a psychodynamic interpretation of the early psychic development of people with learning disabilities, and as such is more difficult to investigate empirically.

However, the core assumptions and beliefs of Gentle Teaching have some areas of overlap with applied behavioural analysis. These include the acceptance that the values which are made explicit by Gentle Teaching should be congruent with those of applied behavioural analysis and the use of the communication metaphor to interpret behaviour. The use of the communication hypothesis has been criticised by some behaviourally oriented researchers, for not distinguishing between messages with functional message value and others. However, the main difference in interpreting behaviours as communication is that a behavioural analysis allows more than one message to be communicated.

The techniques of Gentle Teaching

McGee & Menolascino (1992) present a range of "Supportive Techniques" which they claim can be used to effect the process of mutual change central to the philosophy of Gentle Teaching. These include: errorless teaching; task analysis to simplify tasks and facilitate participation; environmental manipulations; warm helping; co-participation

with the client; the use of tasks to engage the client, whilst maintaining the focus on the relationship; awareness of behavioural precursors; reducing verbal and physical instructions; increase choices available; prompt fading and dialogue. It has been remarked by many behaviourists (Jones & McCaughey, 1992; Bailey, 1992) and acknowledged by McGee & Menolascino that these are largely behavioural techniques.

In a Gentle Teaching approach, however, these techniques are not employed with the aim of increasing or decreasing a specified behaviour or for the purpose of learning a new task or skill. Rather any technique is employed to: “give caregivers the opportunity to express feelings of security, teach the importance of mutual engagement and to deepen the meaning of being valued and valuing others” (McGee & Menolascino, 1992).

A Gentle Teaching approach also seems to eschew any formalised application of these techniques in the form of a programme. Carers are warned against viewing the techniques offered as a “cookbook formula”, rather it is left to the carer to question their values and be continually creative in their application of techniques to produce feelings of security, engagement and valuing (McGee & Menolascino, 1992).

So, whilst applied behavioural analysis and Gentle Teaching may share techniques, it seems that they are used for quite different purposes. In Gentle Teaching the emphasis is on the processes occurring during the interaction, to the extent that the content of the task is immaterial (Golding, 1995). The commitment required from staff is underlined by the responsibility placed upon them to provide the desired conditions.

Strengths of Gentle Teaching

Although some researchers claim that there is nothing positive about a Gentle Teaching approach which is not incorporated into good behavioural interventions, the value of the Gentle Teaching literature may be in making these elements explicit.

Cited by Jones & McCaughey (1992) are the broad overview of a Gentle Teaching approach and the emphasise on mutual change of caregiver and client.

Gentle Teaching is explicitly not about changing individual behaviours. In drawing attention away from set tasks and isolated techniques, and concerning itself with the broader environment of the client, it avoids the client's behaviour being seen in a void and gives a context in which the person is operating. Crucially this includes the carer's own perception of the client and their attitude towards him. Whilst a broader ecological perspective is advocated by many behaviourists (LaVigna & Donnellan, 1986), there is some indication that the field has been slow to adopt this position (Jones & McCaughey, 1992). However, recent papers have begun to examine the wider context in which behaviours occur, examining the emotional effect on carers working with clients with challenging behaviours (Hastings & Remington, 1995) and emphasising the importance of assessing staff beliefs about the maintaining factors of challenging behaviour (Fenwick, 1995).

The acknowledgement that challenging behaviour inevitably impacts on carers, supported by the papers above, lends some support to the assertion of Gentle Teaching that the process of change in a client must be accompanied by change in their carer. However, the nature of change in the carer is difficult to define. According to Bailey (1992), as any behavioural programme requires the carers to do something differently, the process of mutual change is not a unique strength of Gentle Teaching. The levels of change described in a Gentle Teaching approach seem to be far more fundamental than those described by Bailey.

Thus, the main strengths of Gentle Teaching may be that they spell out the need for a broad perspective on a clients behaviour, and emphasise that change in the client requires some change on the part of the carer. It would be of interest to discover if changes in carer behaviour as part of a successful behavioural programme included changes in attitudes towards clients. If so, there would be some empirical support for

the idea that behavioural interventions could bring about changes in staff attitudes as part of the intervention, or whether this can only occur as part of a specifically Gentle Teaching inspired approach.

Difficulties with Gentle Teaching

Three major difficulties with the Gentle Teaching approach will be considered: the validity of its fundamental belief, the misrepresentation of other research and its efficacy.

As stated above, the fundamental belief of Gentle Teaching is that all forms of challenging behaviour are communicative, and arise from abnormal experiences of bonding. This has been contrasted with a functional analysis approach to challenging behaviour which assumes that some behaviours are communicative, but more importantly that they can communicate a range of messages. The rigidity of Gentle Teaching in this respect has led to accusations that it could be dangerous (Jones & McCaughey, 1992), as behaviour arising from any other source (such as an untreated medical complaint) would not be discovered. Of course, any approach can be criticised for not incorporating other models, but the inflexibility of Gentle Teaching makes it particularly vulnerable to this criticism.

A second problem with much of the published work on Gentle Teaching is its attitude towards other research done in the field of challenging behaviour. Reviewing the first published book on Gentle Teaching (McGee, Menolascino & Menousek, 1987), Mudford (1985) concluded: "the ill-researched and vitriolic attack on mainstream behaviour analysts/therapistsis definitely incorrect and possibly libellous". McGee *et al* (1987) were found to misquote and misinterpret behavioural research published by others. Despite continuing criticisms (Jones & McCaughey, 1992), Gentle Teaching literature continues to parody mainstream behavioural analysis, using "culture of death" imagery (McGee & Menolascino, 1991) and providing crude and

inaccurate descriptions of behavioural practices. This serves not only to alienate them from mainstream behavioural analysts, but also questions both the sources of their material, and the accuracy with which they report their own research findings.

Finally, the efficacy of Gentle Teaching should be addressed. The initial problem in determining "efficacy" is defining what should be measured and how success is to be defined. This is hampered by the difficulties outlined above in operationally defining Gentle Teaching procedures. To an extent, this has been alleviated by the development of two instruments, especially to evaluate Gentle Teaching - the CIOS (Caregiver's Interactional Observation System) and the PIOS (Person's Interactional Observation System: McGee & Gonzales, 1990). The generalisability of using a measure designed for Gentle Teaching to evaluate services must be questioned, as it is apparent that most behavioural programmes are evaluated in terms of overt behaviours. Therefore there is the possibility that a programme could be deemed effective in terms of one system, but ineffective in terms of another. Alternatively, if a programme succeeds according to both measures then the discrimination between the two systems must be questioned i.e is Gentle Teaching just "old wine in new bottles" (Cuvo, 1992).

A review of published studies of Gentle Teaching (Jones & McCaughey, 1992) concludes that Gentle Teaching is effective for some clients but not for others. This is consistent with some challenging behaviour being reinforced by social attention, hence implying that Gentle Teaching would be a positive event, with some behaviour being reinforced by other variables such as the removal of task demands or attention. It is for these clients that Gentle Teaching may be an aversive intervention. For McGee & Menolascino (1992), however, this may not be an issue. They state that:

"Doubtless reward and punishment often work. But whether something works or not is not the issue. The central question is what kind of relationship do we want to create."

This is a catch-all, end justifies the means argument which rejects any other than a Gentle Teaching approach even if it has been successful in reducing a challenging behaviour. It also reiterates the belief that the only worthwhile relationship between client and carer is that forged through Gentle Teaching methods.

The relationship between Gentle Teaching and Applied Behavioural Analysis

The preceding discussion has revealed many areas of common ground between Gentle Teaching and applied behavioural analysis. Indeed, when Gentle Teaching was first introduced as an approach mainstream behaviourists had no difficulty in classifying it as either a non-intrusive “Level 1” procedure (Mudford, 1985) or as a variation of stimulus control techniques (LaVigna & Donnellan, 1986). Since then, Gentle Teaching has striven to assert its individuality, and some behaviourists have distanced themselves from the perceived lack of scientific method in the Gentle Teaching approach (Bailey, 1992).

Although some of the techniques of Gentle Teaching may be similar to behavioural procedures, the rationale for applying them and the way in which they are selected is quite different from an approach based on behavioural analysis. However, as described above, there may be some cases when the assumptions underlying a Gentle Teaching approach are congruent with the function of a behaviour for an individual. In this case, a Gentle Teaching approach may operate in a similar way to a behavioural intervention.

A position which seems to incorporate many behaviourists’ perspective on Gentle Teaching is that the explicit value given to a client, and the emphasis given to the commitment and involvement of the carer can enhance the use of behavioural methods with people with learning disabilities (Golding, 1995). Thus selective application of a

Gentle Teaching approach could prevent behavioural techniques being decontextualised, and ensure that clients were treated with unconditional value whatever their level of challenging behaviour.

In conclusion, a Gentle Teaching approach seriously limits the range of interpretations of a client's behaviour, such that it would not be the treatment of choice for all challenging behaviours. There may be occasions where Gentle Teaching and behavioural analysis would come to the same end-point, by applying the same sort of techniques, but with a different rationale underlying each procedure. The main contribution which Gentle Teaching has to make to services for people with learning disabilities is a reaffirmation of the values which should underpin interventions.

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Child and Adolescent Essay

Does cognitive-behaviour therapy work for children and adolescents?

Assessment A17

Cognitive - behavioural therapeutic approaches have become a popular intervention strategy used with child and adolescent psychological problems (Gabor & Ing, 1991). Unfortunately, this is no guarantee of efficacy, and even if efficacy is demonstrated it in turn does not explain the cognitive processes presumed to underlie the psychological pathology and mediate change (Powell & Oei, 1991).

This essay will examine the nature of childhood disorders and the rationale of applying cognitive techniques to various disorders of childhood. Intervention should be based on a coherent cognitive model of a given disorder which ideally would be specific enough for individual techniques to be applied and evaluated. The available research on these issues will be discussed.

The nature of childhood disorders and their relationship to forms of cognitive - behavioural therapy

There are two broad cognitive behavioural approaches which are currently applied to childhood disorders (Kendall, 1985). One of these is Self-Instructional Training (S.I.T.; Meichenbaum 1977), which was derived from empirical work on language acquisition by Luria (1961); the second, clinically derived theories such as Beckian cognitive behaviour therapy (C.B.T.; Beck, 1976) and rational emotive therapy (R.E.T.; Ellis 1967) which were originally derived from work with adults and have later been modified for use with children and adolescents. Thus, if childhood disorders can be conceptualised within the same framework as adult disorders it would seem reasonable to apply the same psychological therapeutic strategies. If childhood disorders, however, are seen as discontinuous with adult, then there is no empirical basis for believing that the same strategies should be effective and novel therapeutic approaches need to be developed.

The conceptualisation of child and adolescent disorders, as exemplified in current classification systems (e.g. DSM - IV) (APA, 1994), allows for both models of disorder. Thus there is a group of disorders which are conceptualised as limited to childhood onset and to be separate from adult disorders although they may persist into adulthood. This group includes Developmental Disorders, Disruptive Behaviour Disorders of Childhood and Anxiety Disorders of Childhood and Adolescence. Otherwise, the diagnostic criteria for adults are thought to be applicable across the age range (Gelfand & Peterson, 1985), so those criteria which define "Major Depressive Disorder" in adults can be used with children (Kaplan & Saddock, 1990).

This scheme is not absolute and reflects the current state of knowledge of childhood psychological problems. However, accepting this model allows for empirical testing of models which in turn can inform future research. It does not explain why childhood disorders can be conceptualised in this way, nor does it give information on the validity and reliability of using this system. Work by Achenbach (1982) suggests that childhood disorders have an underlying three factor structure: two main groups of disorders emerge under the broad headings of "internalising" and "externalising" disorders, with a smaller separate group labelled "pathological detachment". The internalising group subsumes DSM - IV disorders such as anorexia nervosa, the externalising group pathology such as conduct disorders. These three factors seem robust, and have been replicated (Gelfand & Peterson, 1985). Research has also indicated that there may be a similar underlying three factor structure to adult disorders (Eysenck, 1960). Although it is tempting to conclude that this may indicate basic affinities between child and adult disorders, there are various complicating factors. First, childhood problems are likely to be determined by a variety of respondent reports (for example, parents' and teachers') in addition to those of the child. Such reports are likely to yield differing data (Achenbach & McConaughy, 1987) and be subject to positive and negative halo effects, that is the emotional state of the respondent will colour their view of the child's behaviour. Secondly, the factors

which emerge from ratings scales are constrained by the variables entered in the first place: the factors may therefore reflect what researchers already think about psychopathological classification. Finally, no study can be exhaustive in the population group studied, and there is always the possibility that the population group chosen to study is biased.

It is unreasonable to expect that one theory or therapeutic approach will be broadly applicable across the range of psychological disorders. If one wants to use cognitive models to understand childhood psychopathology, then it is necessary to integrate academic knowledge of children's cognitive development with empirical work on the nature of childhood disorders. Then, one approach to intervention would be to attempt to understand the cognitive processes which might underlie disorders, and match strategies accordingly. Amongst psychological theories of disorders, only cognitive models explicitly acknowledge that different psychological processes operate at different times in the life span: the same principles of operant and classical conditioning are assumed to underpin behavioural theories, whilst psychoanalytic theories postulate the same intrapsychic mechanisms at work in child and adult disorders (Gelfand & Peterson, 1985).

One scheme for conceptualising childhood psychopathology is to view disorders as arising from either a deficit or distortion of age-appropriate cognitive skills (Kendall, 1985). Thus externalising problems could be seen to arise from a lack of control, mediated by a failure to acquire age-appropriate self-regulatory internal language. This would be seen as a deficit in an expected cognitive skill. Conversely, anorexia nervosa, an internalising disorder, could be seen as resulting from a distortion of normal cognitive processes of perception and self-image (Kendall, 1985). These two basic forms of cognitive pathology (deficiency versus distortion) map neatly onto the two broad schools of cognitive behavioural therapies outlined above. So, externalising disorders would be most sensibly tackled by using S.I.T. techniques which aim to enhance self-control, and overtly provide the self-regulatory talk which is presumed

absent. Those disorders which involve dysfunctional beliefs and assumptions may be more amenable to C.B.T. or R.E.T. techniques.

This distinction is useful, but unfortunately disorders do not fall quite so neatly into maladaptive processes of deficiency and distortion. For example, anger and aggression fall into Achenbach's cluster of externalising disorders, and so could be approached by teaching S.I.T. techniques (Gabor & Ing, 1991). However, children and adolescents who show excessive anger and aggression also show distortions in cognitive processes such as attention and memory (Kendall & Lochman, 1994). Thus further empirical work into the nature of such disorders is required to elucidate the underlying relationship between deficiencies in normal development and distortions in cognitive processing which can both be hypothesised to contribute to the development of a disorder.

Which children can be expected to work in a cognitive - behavioural way?

Cognitive behavioural approaches demand a certain way of working from practitioner and client. The therapist's role is one of "consultant, diagnostician and educator" (Kendall & Lochman, 1994), with the aim of finding dysfunctional cognitions, formulating hypotheses and then testing these out in collaboration with the client. The client needs to be sufficiently cognitively advanced to be able to participate in this active collaborative approach. Therefore, depending on the conceptualisation of the problem, the client will need to have skills such as the ability to self - monitor, to recognise and report on feelings and thoughts and to be able to reflect on their thoughts and appreciate how these mediate their behaviour. The ability to understand and manipulate cognitive processes using these sorts of skills is often termed "metacognition" (Reeve & Brown, 1985).

Piagetian theory of development has been integrated with cognitive behavioural childhood models to predict those children who should have the metacognitive ability

to use cognitive behavioural techniques (Kinney, 1991). As the techniques require a degree of abstraction, perspective taking ability and the logical and empirical analysis of dysfunctional cognitions, it has been hypothesised that only children who are at the formal operational stage of development are able to benefit from cognitive - behavioural techniques. This position is lent some report by findings that cognitive - behavioural techniques are far more effective with children aged 11 to 13 (and presumed to be in the formal operational stage of development) than children aged 5 to 7 or 8 to 11 (Durlak, Fuhrman & Lampman, 1991).

The Piagetian model of children's development has been extensively researched and refined since its original development. Underlying the original theory was a model of development characterised by structural changes which were invariant in order and irreversible. More recent research has shown that children can actually think in a manner representative of several stages depending on the situation (Donaldson, 1978: Gelfand & Peterson, 1985), thus questioning Piaget's global hypothesis of cognitive development.

Another related question is the extent to which the sorts of logical problem solving abilities central to Piagetian theory are the most important in cognitive behavioural work. An ability to take different social perspectives (Kinney, 1991) and to consider the relation between self and others may be more important. Similarly, empirically testing out ideas of right and wrong or fairness would depend on the stage of moral reasoning that a child was capable of (Kohlberg, 1976).

The implication for practising cognitive behavioural therapy is that relying on the age of the child to determine suitability is not sufficient. An assessment of a child's ability to think about the problem is required which includes the logical cognitive processes needed to examine the situation rationally and the ability to understand the problem in relation to self and others. Furthermore, any intervention planned needs to be guided by normative data for that age group. This applies to alternative thoughts and beliefs

generated by the client and also to intervention planned by the therapist. For example, if social skills training were indicated, then the content of the program should be dictated by normative expectation of social skill for that age group (Kendall, 1985).

A related issue is that the problem may arise from a person being stuck at a certain level of cognitive development in relation to the problem. Beckian cognitive behavioural therapists working with adult personality disorders hold that the personality disorder is underpinned by early maladaptive schema. The material in the schema (e.g. memories, beliefs) is still processed in same way as when the schema was formed. Thus, although the person is functioning in an adult way in many domains of their life, emotionally painful material is still being re-experienced in an immature way.

Further research is needed to determine the extent to which a child's cognitive level and development is responsible for the type of disorder presented (Kinney, 1991). As cognitive models hold that biases in thinking are central to the maintenance of disorders, then it follows that a child who shows the cluster of symptoms associated with depression must also be capable of showing the same errors in thinking. If this is not the case then there are few grounds to transplant cognitive models of adult psychopathology onto childhood disorders. So far, little empirical work has examined the types of cognitions found in clinically disordered children (Kendall & Lochman, 1994).

Thus, practising cognitive behavioural therapies with children and adolescents requires an individual assessment of each client's cognitive abilities, but also a knowledge of normative developmental processes and age-appropriate behaviours and skills. Therefore it is important to broaden the remit of the standard cognitive-behavioural model (involving cognition, behaviour and physiology) to include developmental considerations and the importance of affect in conceptualising disorders (Craighead, Meyers & Craighead, 1985). For example, a child who is impulsive may show a deficiency of self-talk, but the affect associated with this (e.g. anger) may prevent the

metacognition necessary to reflect on the process and remediate it (Kendall, 1985). A successful intervention, therefore, would need to improve the child's understanding and awareness of his emotions as well as to teach self-talk.

The previous sections have highlighted some of the theoretical difficulties in using cognitive behavioural interventions with young people. Childhood disorders can be classified into those which are predominantly due to deficits of normal developmental processes and those which are due to dysfunction. These map onto a conceptualisation of childhood disorders as either predominantly externalising or internalising. This dichotomy is imperfect, as disorders can show both types of cognitive pathology, but may be a useful framework to guide intervention.

Is there evidence for the efficacy of cognitive behavioural techniques?

Despite the limitations of cognitive models of childhood disorders, cognitive behavioural techniques have been applied to a wide range of disorders. Two recent reviews (Durlak *et al*, 1991; Powell & Oei, 1991) aim to determine the efficacy of these techniques and the processes underlying change.

Durlak *et al* (1991) investigated three research questions in addition to determining the overall efficacy of cognitive behavioural interventions. These were to see if cognitive level, as indicated by age, should moderate treatment efficacy; if cognitive change was accompanied by behavioural change and to estimate the practical effect size of treatment i.e. how "normal" children became after treatment.

They found a main effect of treatment, indicating that cognitive behavioural techniques were effective for all types and severity of child problems, regardless of the constituent components of treatment. To investigate the effects of age they divided up the studies into three age ranges 11-13, 8-11 and 5-7. The effect of therapy on older children was almost twice that on the younger two age groups, which did not differ from each

other. The correlation between cognitive and behaviour change was investigated by examining studies which had included a measure of each. There was no correlation between changes in cognition and changes in behaviour. Finally the effect size of treatment was computed. Whereas before treatment the average functioning of experimental and comparison groups was significantly different from normal limits of behaviour, at posttreatment the experimental groups were no different from normal, whilst comparison groups remained different from the norm.

Taken as a whole, these findings suggest some support for cognitive behavioural interventions. However, interpreting the results leaves many questions about the efficacy and process of therapy unanswered. Any effect of intervention is modified by many factors including: the nature and severity of the disorder; the appropriateness of treatment chosen; the interaction of these factors; the methodology used to measure change and the characteristics of the children treated. As the analysis took all types of problem and therapy together it gives little information on the whether specific disorders or age groups responded differently to a given approach.

As outlined before, age alone is not a sufficient predictor of suitability for cognitive therapy, so the enhanced efficacy with age should be interpreted with caution. This is especially pertinent as there was no difference in efficacy between the two younger age groups. If suitability for therapy were directly related to cognitive development which is assumed to develop monotonically with age then there should be an increase in efficacy across age groups. The fact that this did not emerge suggests that something else is modifying treatment efficacy for the youngest age group. One possibility is that they were treated by different techniques. If, for example, S.I.T. techniques were used then they might interact with the normal emerging verbal abilities to enhance efficacy.

The relationship between cognitive change and behaviour change was examined using the 33 out of 64 studies which included measures of both these variables. The conclusion was that cognitive change was not related to changes in behaviour. This

finding is limited by the nature of the questionnaires used and their lack of specificity. Measures of cognitive change included the Matching Familiar Figures test (a measure of cognitive tempo) and measures of personality functioning (details not given). It is debatable whether questionnaires which aimed to tap more directly into, say, attributional style or beliefs would have produced more evidence for correlations between cognitive and behaviour change.

Powell & Oei (1991) further investigated evidence for cognitive processes underlying behaviour change in childhood disorders. They identified 63 studies which incorporated a cognitive behavioural approach (again broadly defined to include S.I.T. and C.B.T. approaches). Of these eight combined cognitive behavioural and other approaches and therefore give little information on the efficacy of any given technique. seven studies combined several cognitive behavioural techniques with or without comparison groups. These provide some evidence for the efficacy of a barrage of cognitive behavioural techniques, but little information on the process of change. 15 studies compared cognitive behavioural approaches with other forms of intervention, the results suggesting that cognitive behavioural approaches have some superiority over pharmacotherapy and traditional behavioural techniques.

24 from the original 63 studies tried to isolate and/or compare specific cognitive behavioural techniques. These showed that S.I.T. techniques were successfully used to improve academic ability; reduce shyness; control aggressive behaviour; reduce self-defeating behaviour and increase social skills. Unfortunately the analysis did not divide the studies into ages of children seen, therefore the tentative hypothesis from the Durlak *et al* study of S.I.T. being more suitable for younger children cannot be examined. Other cognitive techniques, such as anger management, also produced positive change.

Only nine out of 63 studies attempted to measure cognitive change, and therefore provide a means of examining the process of change. These studies typically used self-

report, self-concept questionnaires and overall showed correlations between cognitive and behavioural change. However the methodology used was not sufficient to demonstrate that behaviour changed as a result of cognitive change - a basic assumption for improvement in cognitive behaviour therapies.

The difficulty in demonstrating that cognitive change mediates behavioural change is unsurprising, given the range of cognitive factors which are thought to contribute to disorders. At the very least, researchers differentiate cognitive events, processes and structures (Harris, 1985), which can be further divided into expectancies, current concerns, attributions, beliefs and many others. In the main, these are not well-differentiated concepts, which makes devising reliable and valid assessment tools all the harder. The likelihood is that a variety of assessments, carried out at different times will give the most accurate assessment of a disorder (Kendall, 1985). Achenbach & McConaughy (1987) also stress the importance of using multiple raters of behaviour as information from only one source will not provide a whole picture of a disorder.

The battle to prove that cognitive processes underlie and mediate behaviour change is likely to be a long and frustrating one. At the heart of the problem is the lack of specificity of childhood disorders. In addition to identifying cognitive, behavioural and physiological variables associated with disorders it is vitally important to consider the developmental stage of a child and how that will impact on presentation, modify therapeutic strategies and determine appropriate outcomes. Without models which can predict key distortions or dysfunctional styles it is difficult to either devise a reliable and valid measure for measuring change along those dimensions or select a therapeutic strategy which would best modify it.

Special considerations when working with children and adolescents

The previous review papers show that, despite having few grounds for specifying that cognitive processes underlie behaviour change, cognitive behavioural techniques can be usefully employed across a wide range of ages and disorders. Given the previous discussion of the lack of specificity of models, and the many problems associated with selecting the most appropriate treatment strategies for a given disorder it seems surprising that there is any beneficial effect at all. This last section will consider particular problems and benefits of employing cognitive behavioural techniques with children and adolescents.

An issue frequently raised by researchers is that children and adolescents may be unaware of or unwilling to admit that there is a problem and seldom self-refer (Gabor & Ing, 1991; Powell & Oei, 1991). Their participation in therapy may be as a result of wanting to please or pacify a third party (such as a parent or teacher) or to get a secondary gain such as time away from school. Thus the first task of the therapist is to assess the child's motivation for therapy, and their reasons for participation. The formation of a working alliance is central to the practice of any form of cognitive behavioural therapy.

Secondly, the pressure to conform may be greater for children, and this may lead to an overestimation of the efficacy of cognitive techniques to produce changes in beliefs and attitudes. Children may not have the ability to rationally examine and change their thoughts and beliefs, but they may be able to infer what the therapist would like them to say. In literature which reports the use of positive self-statements with young children (e.g. Gabor & Ing, 1991) it is unclear whether the more directive style advocated for use with younger children because of their limited ability to generate other ideas for themselves can become lecturing.

However, there are positive aspects of using cognitive behavioural techniques with adolescents. McAdam (1986) points out that the active style of therapy may suit work with adolescents, by avoiding uncomfortable silences and presenting therapy as a collaborative effort. Conversely, there may be many teenagers who are quite perturbed by a continually questioning and summarising therapist, who could easily be perceived as intrusive.

Conclusion

Cognitive behavioural techniques, defined as either those which emerged from empirical work on children's language acquisition or those which have been scaled down from adult models of psychopathology, have been shown to be effective with children and adolescents. However the lack of specificity of cognitive models of disorders (Powell & Oei, 1991; Kendall, 1985) hampers research into the cause and maintenance of disorders and the development and application of the best interventions.

The best assessment and intervention incorporates developmental research and takes account of normal cognitive processes and behaviour for a given child. The importance of broadening standard cognitive behavioural models to include developmental factors cannot be overemphasised (Kendall, 1985). To assess a given child it is important to take multiple measures of behaviour (Achenbach & McConaughy, 1987) from different raters and at different times (Kendall, 1985; Harris, 1985). In intervention, practitioners should be aware of the difficulties of working with problems which have not been self-referred and maintain a collaborative, rather than authoritarian stance. This may be especially hard when working with younger age - groups in which it may be necessary to be more didactic.

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Older Adults Essay

**Discuss the issues and concerns in the provision
of group therapy for older people**

Assessment A19

“near or above the fifties, the elasticity of mental processes ...is as a rule lacking - old people are no longer educable.”

Freud, 1905.

Contrary to Freud's position, researchers are increasingly interested in the changes which can be brought about by working psychotherapeutically with older people. This includes research into the possible advantages of using groupwork with this population. An important component when considering the issues and concerns of groupwork with older adults is to identify areas of communality between this population and others. As any dividing line between the “old” and the “general” population is arbitrary, it is not to be expected that there would be a sharp, quantitative difference between the two groups in the content and process of groupwork.

The older adult population is a homogeneous one, thus the issues and concerns in providing psychological services vary with the sub-group under discussion. The population can be divided along various lines, such as sex, physical disability, age range and so on, some of which will be more pertinent to the issues of groupwork than others. A salient dimension along which to divide the population is to consider the provision of groupwork with the cognitively intact separately from organically impaired older people. As will be discussed, the needs of, and methods applicable to the two groups are different.

Some aspects of groupwork are age-independent

The areas which are common to groups regardless of age are those concerned with process rather than content. As will be discussed further, the themes and areas of concern to older people tend to be different from younger adults (Hildebrand, 1982). Process variables are concerned with the formation and development of a group: delineating a model of normal or typical group processes for older adults should facilitate research into group work with this population.

A) Models of group process

Bender (1990), using a general model of group processes which hypothesises a take-off and dissolution phase of group development, proposed that group member involvement as measured by the Involvement Scale (Bender, Norris & Baukham, 1987) is a variable which correlates with the development of the group. During the initial group sessions the over-all involvement of members will increase, and as the end of the group approaches the members' involvement will decrease. Bender & Morris (1990) report that this pattern is true of groups consisting of older people with dementia as well as the cognitively intact, implying that the process of group development is relatively constant across populations.

There are a number of factors, however, which may hamper the normal process of development in groups of older people. First, members may have physical health problems which impede their attendance or involvement on a session to session basis. Related to this point is that this effect will be disproportionately large if ill-health affects a prominent group member. In addition, this factor may be more salient than the group development effect. Thus the overall pattern of group development may only emerge from an averaging of the processes from a number of groups. Finally, the enthusiasm and input from group leaders may be a major factor in determining the continuing work of the group: that is, a loss of enthusiasm by the leaders may lead to a premature decline in group involvement.

The tentative conclusion is that one should assume a model of group development which is similar to that in other age-groups, and that this development process is the same for both cognitively intact and organically impaired groups of older people. This process may be disproportionately affected by the actions or health of individual group members or leaders.

B) Curative factors

The second area of communality between groups for older people and other age groups is that the curative factors in group therapy are not age-constrained (Tross &

Blum, 1988). There are various models and numbers of curative factors hypothesised in groupwork (e.g. Yalom, 1985), and there is substantial agreement as to what these factors are and the number hypothesised (Bender, 1990). These curative factors are vital to the success of groupwork with older adults (Leszcz, 1990; Sorensen, 1986) but this population group may have unique difficulties in achieving an environment in which these factors can emerge.

For example, one of the most important curative factors is cohesion, which Leszcz (1990) describes as the “backbone of treatment”. It refers to the creation of a group environment in which members feel understood, accepted and valued, and can begin to re-engage with their environment (Sorensen, 1986). However, there are a number of factors which are applicable to older people which may impede the formation of group cohesion. Cohesion forms and develops as members become aware of areas of overlap and communality in their personal and societal history (Sorensen, 1986). This is vulnerable to being impeded by the characteristics of the members. For example if sensory loss or communicative problems exist (Hunter, 1989), members may have real difficulty in establishing meaningful relationships. The behaviour and attitude of the therapist can also hamper the development of a group where curative factors can emerge. This can be either by not working actively to promote interactions from which relationships can develop (Hunter, 1989) or by implicitly devaluing the group. Sorensen (1986) considers that the commitment shown to the group by the therapist (which is shown through basic principles such as the maintenance of boundaries, and other gate-keeping functions) is an important factor in indicating to the group that they are valued and important. A negative countertransference from the therapist to the group will be met by resistance by the group, which will act against cohesion developing.

Thus, although the curative factors in groupwork with older people are hypothesised to be the same as in work with other age groups, the factors which may hinder their operation can be from a number of sources. First, the physical or mental attributes of the group members may make participation in the group difficult, and act to hinder the development of curative factors such as cohesion, inclusion and trust. Secondly, the

role of the therapist may be more important when working with older age groups to ensure that a therapeutic environment is formed and maintained. This may require the therapist to be active in promoting the individual participation of members, or ensuring that the impact of physical or sensory disabilities are minimised to promote communication and the formation of relationships within the group. Finally, it is the responsibility of the therapist to recognise and act upon their own negative transference towards the group, and towards older people in general. The problem of negative countertransference developing may be particularly difficult for therapists working with older people; almost invariably the older people encountered will be those who are having difficulties in coping with the changes caused by ageing (Bender *et al*, 1987). The opportunities for maintaining a balanced picture of the ageing process may be limited.

The research suggests that the process of group development and the curative factors in group work are robust, regardless of age. The issues of concern in applying these models to working with older adults is to ensure that the conditions under which the groups are run, and the training and awareness of the therapist are optimal for the process to develop and the curative factors to emerge. Thus, for example, the therapist may need to take a more active role in stimulating those group members for whom communication is difficult, or in arranging the group so that disabled members can most easily take part. Individual actions and problems may have a disproportionate effect on the group process.

The content of groups may be unique to older adults

Leszcz (1990), in his paper arguing for an integration of different group models for use with cognitively intact older people, describes three different types of psychotherapeutic groups: psychodynamic, developmental and cognitive behavioural. The first two, being concerned with life-long psychological development, hypothesise unique issues for older people, which need to be dealt with.

A: Age related psychodynamic concerns

Hildebrand (1990) describes five major changes which occur with age, and which may lead to problems in later life. The first is that of loss, whether of external objects and significant others or from sensory or physical disabilities. Society can also play a part in promoting ageist attitudes and images, which can adversely influence an older person's self-image. These losses can undermine the "healthy narcissism" of an individual, and lead to a withdrawal from everyday life in order to protect oneself from further loss (Sorensen, 1986). Secondly, ageing can highlight changes in object relations (such as between spouses, or children and parents) which need to be addressed. The next two factors concern changes in gender roles. According to Gutmann (1977), men tend to be characterised by a shift from agency to community, whereas women age psychologically in the opposite direction. Difficulties may arise, then, if a man has difficulty in accepting and assimilating his new feminine side or a woman is troubled by the emergence of aggressive or competitive urges. Finally, the expression of sexuality may be important to older people, especially as society tends not to recognise or condone sexual expression in older people, despite the evidence that sexuality remains important throughout the lifespan.

For these psychodynamic changes in later life, group therapy may well be the treatment of choice in promoting resolution and acceptance. Sorensen (1986) describes how the curative factors of cohesion, mastery and inclusion can act to aid adaptation to losses in later life. This can be by halting the cycle of withdrawal which may be precipitated by loss, and providing a valued environment in which an individual can begin to re-establish contact with the environment, and regain a sense of their own mastery through the formation of new relationships. A group can provide a supportive environment in which one's opinions and experience are valued, and can help the older adult regain a realistic sense of self. Finally, a group can provide an environment in which changes can be normalised, or on-going behaviour accepted.

B: A developmental model

The second type of groupwork which is commonly used with older people is reminiscence. This is linked to a developmental model in which a life review is both normal and desirable, and can be used to re-establish a sense of who someone is, by linking with who they were (Leszcz, 1990). A concern when adopting reminiscence techniques with older people is that reminiscence can be antitherapeutic as well as beneficial. For cognitively intact people, reminiscence can help to resolve conflicts and reintegrate these events into their personality. For example, in reminiscing, a person may encounter memories of events which were not resolved at the time. With greater age and maturity they may be able to take a different perspective on them and arrive at a resolution. Reminiscence about past important roles or achievements may also serve to enhance self- and public esteem. By recalling events of personal triumphs and accomplishment, an individual may be provided with a sense of importance or self-worth. A further function is that reminiscence may promote re-engagement in preferred activities. Finally, reminiscence may be instrumental in working through loss and grief (Poulton & Strassberg, 1986).

However, it is also recognised that reminiscence can have maladaptive consequences. Persistent reminiscence may constitute an avoidance of here-and-now issues, and in fact be active withdrawal from finding solutions to current difficulties. Reminiscence may also raise memories or issues which an individual is unable to resolve, and thus cause distress. Finally, if not contained appropriately, reminiscence may serve to isolate the individual by alienating other group members.

Using reminiscence therapeutically, which implies creating an environment in which curative factors can develop, requires the therapist to actively promote helpful and adaptive reminiscence. There are various methods for doing this. First, the therapist should be adept at exploring memories, in particular fleshing them out to provide more detail. The quality of a reminiscence determines its therapeutic value (Poulton & Strassberg, 1986). A further skill required is that of linking the reminiscence with

here-and-now concerns, by clarifying the meaning of the reminiscence (such as skill retrieval, resolution of conflict) and drawing the implication for a current problem. To prevent the use of reminiscence to avoid current issues or divide the group, the therapist must be able to switch the group from reminiscence to here-and-now concerns. Finally the therapist may also have to act to promote interpersonal learning from reminiscence material. This implies both making one person's reminiscence relevant to other people, and also ensuring that the group is free from unfair criticism or hostility towards members who bring unpleasant or difficult material to the group.

The therapeutic use of reminiscence has been described above as the attempt to integrate forgotten or unresolved issues from the past to the here-and-now. These aims are usually applied to cognitively intact older people. However, reminiscence can also be used in groups with organically impaired older adults, where the aims are usually more limited. The minimum aim for using reminiscence with organically impaired older adults is to provide a meaningful and appropriate stimulating activity. McKiernan & Yardley (1990) demonstrate that reminiscence groupwork is effective in engaging severely demented clients and enabling them to contribute to complex verbal group activity. Head, Portnoy & Woods (1990) set the aims for their groups as engagement; provision of alternative activity; to encourage communication between group members and to increase the involvement of members as the group progressed. In evaluation of the groups they conclude that reminiscence is only effective in promoting involvement of group members in settings where the opportunities for meaningful interaction were limited. An interesting function that the group may serve is to promote staff-member interactions, in those settings where staff were unclear of their relationship to the group members. This could operate through a number of mechanisms, for example the group being a place where good communication practices were modelled. However, the group was no more successful than other activities in promoting peer interactions. Head *et al* (1990) concluded that the aim may therefore be unrealistic.

In conclusion, reminiscence work with organically impaired older people at the least offers a meaningful activity. The act of reminiscence also capitalises on residual

memory and hence gives the older person an opportunity to show a greater degree of ability than is evident in here-and-now encounters. For example, they may be able to model how to use old items (such as a gas mask) but be unable to name it, or may be able to talk at length about their childhood whilst being disorientated day to day (Bender *et al*, 1987).

Successful use of reminiscence groups with this population raises the following issues. First, it is likely that the group will require a high staff to members ratio, typically one therapist to two group members. The length of groups needs to be kept short, with 30 to 40 minutes being recommended (Bender *et al*, 1987). The therapist may have to alter their style of communication to facilitate understanding, so sentences need to be kept short, to avoid ambiguity and to pose a specific question (Sorensen, 1986). Finally, the material of the group may need to be simplified for use, but this should not mean that it becomes patronising (Bender *et al*, 1987).

The final concern in using reminiscence groupwork with elderly clients is that the research into its efficacy is equivocal. As Poulton & Strassberg (1986) point out, this is partly because reminiscence is a complex cognitive activity, and it is unclear which variables should be measured or indicators of success. There is little agreement between studies which have attempted to examine function or effectiveness (Thornton & Brothie, 1987). It should be noted that the adaptive functions of reminiscence proposed by Poulton & Strassberg (1986) are a curious mix of functions which would serve to enhance disengagement from personal and social activities by advocating introversion of thoughts and those which promote engagement and the recommencement of activities. It is unclear how successful reminiscence can be at promoting these two diverse aims, especially in the context of groupwork which aims, by its nature, to promote interaction and re-engagement.

The final broad category of groupwork delineated by Leszcz (1990) are those based on cognitive behavioural principles. This differs in its aims from the previous two models by emphasising conscious cognition and learning from examining the cognitions which are thought to maintain the problem. Cognitive behavioural techniques are most

frequently applied to discrete problems, usually depression. In contrast to reminiscence, research shows favourable outcomes for cognitive behavioural group therapy in reducing observer and self-rated depression (Steuer, Mintz, Mammen, Hill, Jarvik, McCarley, Motoike & Rosen, 1994), improvements in cognition (Abraham, Neundorfer & Currie, 1992) and reducing hospitalisation (Weiner, 1992).

As Leszcz points out, an appropriate model for working with the cognitively intact elderly might be to incorporate elements of each of the above approaches to enhance each other. For example, the cognitive technique of positive reframing of an event may be facilitated by asking a person to draw on their experiences to remember a time when they would have seen a problem differently. Engaging a member in the therapeutic alliance may be facilitated by appreciation of psychodynamic concerns, such as the importance of the older person maintaining a sense of mastery rather than feeling cajoled into group participation.

Conclusion

A number of models of groupwork have been proposed for use with the older population. There is a degree of consensus that the processes and curative factors of groupwork are the same for this population as any other, but that problems which are most prevalent in this group (such as ill-health) may require recognition and input from the therapist if the correct therapeutic environment is to be established.

The content of groupwork, from a psychodynamic perspective, is different from other age groups, with losses which represent a significant threat to the healthy narcissism of the individual being especially important to work through. Another model of groupwork which is most often applied to older adults is that of reminiscence, and the importance of life review. Although empirical evidence is lacking, researchers hypothesise a number of healthy functions of reminiscing. However, therapists using a reminiscence model should be aware of the antitherapeutic functions which reminiscence can have. Unlike other forms of groupwork, reminiscence is also used successfully with organically impaired older adults, although the aims of these groups

are often more simple. Cognitive behavioural groups have been shown to be successful with delineated problems within the older adult population, such as depression. However, such an approach may not be the most useful for problems which are without a clear focus. In such cases, an eclectic approach incorporating elements of each model may be more appropriate.

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Neuropsychology Essay

**The contribution of clinical neuropsychology to
the assessment of fragile X syndrome**

Assessment A23

Clinical neuropsychology is concerned with the scientific observation and measurement of behaviours and cognitive processes, with a primary aim of making inferences about brain function (Fennell & Bauer, 1989). Within clinical neuropsychology, paediatric neuropsychology is emerging as a distinct sub-discipline (Griffiths, 1996). It is concerned with the effect of disorders of the central nervous system on normal cognitive and behavioural development. Within this framework two broad categories of disorder can be distinguished: neuropathological and neurodevelopmental. The former includes all disorders whose organic aetiology is known, and of which something is understood about their neurological basis. Fragile X syndrome is a neuropathological disorder. In contrast, neurodevelopmental disorders are those where an organic pathology is hypothesised, but not known. This category includes developmental dyslexia and attention deficit disorder (Griffiths, 1996).

Paediatric neuropsychology has a rather different remit to adult clinical neuropsychology. In addition to sharing the aim of illuminating brain-behaviour relationships, there are a number of other important areas to which a paediatric neuropsychology assessment can contribute. These can be divided into those which are important to the individual and family, and those which contribute to the wider understanding of neuropsychological development.

First, in the case of neurodevelopmental disorders, a neuropsychology assessment may be the main way of making a systematic assessment and diagnosis of the difficulty. In the case of neuropathological disorders, a neuropsychological assessment may be invaluable in making sense of a genetic diagnosis. Secondly, parents' and families' understanding of the nature and implications of a diagnosis of a developmental disorder is crucial to their acceptance of the child, and to the instigation of appropriate interventions (Turk, 1996). Neuropsychology may have a role in this process, as careful assessment of the individual child is important in informing behavioural interventions, and may be important in predicting future development (Bennetto & Pennington, 1996). Thirdly, medication may form an important part of intervention, especially in the case of children with neurodevelopmental disorders. Well-chosen psychological measures could be used to monitor the effects of treatment, and hence

provide an objective aid to decide the most appropriate level of medication (Griffiths, 1996).

Paediatric neuropsychology also has important contributions to make to the wider understanding of neuropsychological development. Whilst clinical interventions are individually tailored, there is increasing information that certain behaviours and cognitive processes may be syndrome specific (Turk & Hill, 1995). These factors should be taken into account when designing a clinical intervention. Similarly, educational strategies can be tailored to the patterns of cognitive abilities shown by children with chronic developmental problems. Finally, paediatric neuropsychology has a vital role to play in the reliable and valid measurement of behaviours and cognitive processes, which in turn may help to illuminate brain-behaviour relationships.

The potential of paediatric neuropsychology to contribute to the assessment and management of children with fragile X syndrome in the above areas will be reviewed. This will necessarily include an appraisal of the current limitations and difficulties in the field.

Fragile X syndrome

Fragile X syndrome is a neuropathological disorder and the most prevalent known inherited cause of learning disabilities. It accounts for around 10% of severe learning disabilities in males with learning disabilities of previously unknown origin (Turk, 1992) and up to 30% of all cases of X-linked learning disabilities (Hagerman, 1992). Fragile X syndrome is the result of a genetic abnormality which adversely affects development, resulting in genetic, physical and psychological factors which characterise people with the syndrome.

The genetic basis of the syndrome is complex. The gene responsible for causing the syndrome is the fragile X mental retardation gene (FMR 1), found on the long arm of the X chromosome. In fragile X syndrome the number of CGG repeats on the FMR 1 gene expands beyond the normal range. In the normal population there are between 6 to 54 CGG repeats (Fu, Kuhl, Pizzuti, Sutcliffe, Richards, Verkerk, Holden, Fenwick,

Warren, Oostra, Nelson & Caskey, 1991), in carriers of the fragile X syndrome this increases to up to 200 repeats. Affected individuals with fragile X syndrome have from 200 to more than 1000 CGG repeats. In addition, there may be methylation of the neighbouring CpG island (Oberle, Rousseau, Heitz, Kretz, Devys, Hanauer, Boue, Bertheas & Mandel, 1991). The effect of the CGG expansion, and associated methylation is to prevent expression of the FMR 1 gene leading to a null mutation in which no gene product is formed. The mechanism by which the lack of this protein product gives rise to the clinical features of fragile X syndrome is not known.

The complexity of the genetic basis of fragile X syndrome means that there are a number of genetic factors which are important in the assessment of the neuropsychological sequelae of the syndrome (Bennetto & Pennington, 1996). For females these factors include: the length of CGG expansion; the degree of methylation and the X inactivation-ratio. Females affected by fragile X syndrome will have one normal and one fragile X chromosome. Their clinical presentation will depend on the proportion of fragile X chromosomes which are active. The important factors for males are somewhat different. They tend to have the full mutation (i.e. in excess of 200 CGG repeats) although the exact expansion of CGG base pairs may be important in determining the degree of methylation of the neighbouring CpG island, which in turn regulates the amount of FMR 1 protein produced. Important genetic factors in their assessment, therefore, are: length of CGG expansion; degree of methylation, and whether they have some cells with the full mutation and some with the pre-mutation (termed mosaicism) (Hagerman, 1996).

Clinically, fragile X syndrome is important for a number of reasons, including explaining the aggregation of learning disabilities in families and the observation that more males than females are affected by learning disabilities (Turk, 1992). It has also attracted a lot of interest as individuals affected by fragile X syndrome tend to show a characteristic pattern of behaviours. Notably these include abnormalities in social interaction, which are behaviourally similar to those seen in autism (Turk, 1992). Initially it was thought that fragile X syndrome may be an major genetic cause of

autism (Hagerman, 1992), with a current estimate that 2.5% of people with autism also have the fragile X anomaly (Bailey, Phillips & Rutter, 1996).

So, in assessing fragile X syndrome a number of important factors must be considered. Some of these are concerned with the accurate identification of the genetic parameters of the syndrome and then linking those to observable behaviours. Others are concerned with the reliable and valid measurement of behaviours and cognitive processes, and using this information to inform interventions and further research into the syndrome. However, there are significant methodological and conceptual difficulties associated with assessing neuropathological disorders, and these will be addressed below.

Conceptual and methodological issues in the assessment of neurodevelopmental disorders

Lack of appropriate models.

First, our current understanding of normal neuropsychological development is limited (Bennetto & Pennington, 1996). During embryonic development, infancy and early childhood the child's brain is developing extremely rapidly (Beardsworth & Harding, 1996). It is more vulnerable to physical, biochemical or viral attacks than mature brains, but also more capable of adaptation and reorganisation if compromised (Griffiths, 1996). Normal models of childhood cognitive processes such as memory and executive functions are largely informed in the first instance by adult models of functioning, although there is recognition that these may not be appropriate models for use with the developing brain (Pennington & Ozonoff, 1996).

A further difficulty is that paediatric neuropsychology commonly uses norm-referenced tests to ascertain group differences on measures of cognition or behaviour. Studies which assess individuals with fragile X syndrome, a condition characterised by a life-long deviation from normal development, on measures which have been standardised and devised for use with normal populations must use appropriate comparison groups

in order to illustrate that patterns of functioning are indeed the result of a developmental deviance and not the non-specific result of developmental delay (Bennetto & Pennington, 1996). Ideally, studies investigating the neuropsychological characteristics of fragile X syndrome would include comparison group of children with learning disabilities of unknown aetiology (to control for the general non-specific effects of having a developmental disability) and another group with a syndromal cause of their learning disability (e.g. Down's Syndrome) to control for the effects of developmental deviance.

Lack of appropriate measures

A related problem is the methodology used to delineate the behaviours characteristic of fragile X syndrome. This issue can be addressed in two ways. Standardised and valid measures of existing syndromes (such as autism) can be used to investigate similar behaviours in other conditions. Alternatively an attempt can be made to assess behaviours which do not fit into an existing diagnostic category by developing individualised measures (Flint, 1996). There are obvious advantages and disadvantages to each approach. Standardised measures provide a yard-stick against which to compare the population under test, but in the case of fragile X syndrome this has led to the erroneous belief that individuals with fragile X syndrome were more likely to have autism than others of the same intellectual ability. Many of the behaviours associated with fragile X syndrome (such as poor eye contact and hand flapping) are indeed similar to behaviours common in people with autism, and studies have shown that some groups of people with fragile X syndrome have elevated levels of behaviours associated with autism compared to comparison groups (Kerby & Dawson, 1994). However, it cannot be inferred that the same underlying cognitive or biological processes mediate similar behaviours (Fletcher & Taylor, 1984). Indeed, when non-standardised measures of more specific aspects of behaviour are used important differences between fragile X syndrome and autism emerge. For example, boys with fragile X syndrome have been found to be sensitive to parent-initiated gaze, and to show gaze aversion unrelated to their verbal ability or age in contrast to boys

with autism (Cohen, Vietxe, Sudhalter, Jenkins & Brown, 1989). If standardised measures alone are used, then such subtle differences will be missed.

The complex nature of fragile X syndrome

Finally, the genetic complexity of fragile X syndrome complicates any large-scale investigation into reliable behavioural or cognitive features associated with the syndrome. As noted above, groups should be matched on the features of the genotype (such as length of CGG repeat) as well as other parameters such as overall level of intellectual functioning, social class and presence of other psychopathology (such as autism). Although fragile X syndrome is the most common inherited cause of learning disabilities it is still relatively rare, therefore identifying carefully controlled cohorts for study is difficult. However, unless such studies are undertaken, any syndrome-specific behaviour or cognitive process is unlikely to be detected as it will be masked by other genetic and environmental differences between groups (Bennetto & Pennington, 1996).

These are some of the precautions which must be taken in assessing children with fragile X syndrome. The contribution of paediatric neuropsychology to the assessment of fragile X syndrome will be considered with these issues in mind, and with reference to the general aims of paediatric neuropsychology outlined previously.

Contribution of paediatric neuropsychology to the assessment of fragile X syndrome

1) Accurate information for family and other agencies

When a child is identified as having a developmental disability, it is important to provide the family and other agencies involved with accurate assessments of the child's abilities. A neuropsychological assessment is therefore important in providing information on overall levels of functioning and on specific cognitive deficits which the individual might have. It provides a context in which to interpret other assessments (such as of speech and language skills), to determine whether they are consistent with overall levels of functioning. Furthermore, when taken in conjunction with genetic

information, neuropsychological assessments can be important in predicting the course of intellectual functioning. Early studies suggested that males with fragile X syndrome suffered a dramatic decline in IQ at some time between the ages of six and 15. Methodologically sound studies (using longitudinal designs, equivalent measures at times one and two and including genotype information about their fragile X syndrome groups) have found that the decline may be limited to males with over 50% methylation of the CpG island, when no gene product is formed (Wright-Talamante, Cheema, Riddle, Luckey, Taylor & Hagerman, 1996). Further neuropsychological studies need to be undertaken to establish whether the decline is an artefact of the tests used, due to a decline in certain areas of cognitive functioning relative to others (Hodapp, Dykens, Ort, Zelinsky & Leckman, 1991), or due to the failure to develop in certain cognitive areas.

II) Informing clinical and educational interventions

In addition to providing invaluable information to parents and carers on the likely course of their child's intellectual development, careful neuropsychological assessment is necessary to inform clinical and educational interventions. Although certain behaviours may be reliably associated with a syndrome, and are therefore assumed to be partially genetically determined, there is strong evidence that such syndrome-specific behaviours can be controlled by careful behaviour modification programmes (Holland, & Murphy, 1990). This could be in a number of ways. For example, neuropsychological methods could be used to investigate the meaning or function of a behaviour. Males with fragile X syndrome tend to show a range of social behaviours which are similar to those seen in autism. Neuropsychology has a role to play in establishing whether similar cognitive processes (for example, impairments in theory of mind) underlie similar behaviours. Secondly, neuropsychological investigations have suggested profiles of cognitive strengths and weaknesses of carriers and individuals with fragile X syndrome. For example, female carriers of fragile X syndrome tend to have a specific weakness in arithmetic, which may in turn be related to difficulties in executive functions (Mazzocco, Pennington & Hagerman, 1993). Therefore, females with fragile X syndrome should receive support in this area. Finally, there is evidence

that some of the weaknesses characteristic of fragile X syndrome may be due to difficulties in sequential processing of information, with relative preservation of simultaneous processing skills (Dykens, Hodapp & Leckman, 1987). This has clear implications for the most appropriate methods for presenting information to children with fragile X syndrome, and these findings influence current educational strategies for these students (Dykens, Hodapp & Leckman, 1994). Techniques which capitalise on simultaneous processes (such as matching words to objects in the school environment, rather than presenting lists of words to learn) may be most effective for students with fragile X syndrome.

III) Monitoring the effects of medication

It has been mentioned that neuropsychology may have a role to play in monitoring medication, in terms of its effect on cognitive processes. Fragile X syndrome commonly coexists with deficits in attention (Turk, 1992) which may be treated with stimulant medication. However, common unwanted side-effects include over-selective attention and behavioural rigidity (Griffiths, 1996). Although few standardised tools exist to assess such features, the use of measures of attention (such as the Continuous Performance Task) (Hinton, Halperin, Dobkin, Ding, Brown & Miezieski, 1995) could be developed to monitor drug response.

IV) Making inferences about brain-behaviour relationships

It has been stated that the primary aim of neuropsychology is to make inferences about brain-behaviour relationships. There are serious methodological and conceptual problems in doing this in the case of fragile X syndrome. First, the genetic and environmental variability between individuals may mask any effect of fragile X syndrome unless groups are very carefully selected and many intellectual and environmental factors controlled for. This enterprise is further complicated by the number of genetic factors which must also be accounted for in any study. Secondly, current knowledge of normal neuropsychological development is limited, making the relationship between research into neuropathological disorders and normal

development dialectic and reciprocal (Bennetto & Pennington, 1996). This impacts on the development of standardised, norm-referenced tests of a range of cognitive processes in children. At the moment these tend to be based on adult models of functioning which may not be appropriate (Pennington & Ozonoff, 1996). Finally, the assumption that the FMR 1 gene has a specific effect on behaviour may be misguided (Flint, 1996). People with fragile X syndrome have such a range of impairments that it is likely that the FMR 1 gene has a non-specific effect on behaviour by controlling some basic process of cell biology.

Conclusion

Paediatric neuropsychology is an emerging discipline which has a number of important roles to play in the assessment of neuropathological disorders. Its utility in the assessment and treatment of children affected by fragile X syndrome has been described, in the light of current limitations in methodology and knowledge base. It is clear that a thorough and careful neuropsychological assessment, in conjunction with accurate genetic information, can provide important information on the likely course of intellectual development; inform behavioural interventions and help to develop effective educational techniques. Future roles may include greater participation in monitoring medication. The complex nature of fragile X syndrome, coupled with the probable lack of specificity of the FMR 1 gene may limit the inferences which can be made about specific brain-behaviour relationships. However, such inferences rely on the careful and systematic delineation of behaviours associated with the syndrome, an area which is clearly within the remit of clinical neuropsychology.

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Specialist Essay

**Siblings of children with learning disabilities:
recent research findings and clinical implications**

Assessment A31

The birth of a child with learning disabilities has a profound impact on his family, and can be considered to alter the whole family environment, above and beyond the adjustments required when any child is born (McHale & Gamble, 1989). Services are becoming aware of the multiple challenges to families made by children with learning disabilities, which occur across many domains. These may include social, educational, psychiatric and behavioural implications of their child's learning disabilities (Turk, 1996). In many cases, the reaction of a family to the arrival of a child with learning disabilities may be considered in terms of a chronic grief or bereavement reaction, in which the family members have to pass through stages of shock, denial and anger before reaching a stage of acceptance of the child's limitations, and then being able to plan meaningfully for the family's future (Turk, 1996).

Within the context of this emerging awareness of families' needs, the special needs of siblings are being recognised. Clinical services are beginning to offer interventions to siblings to help them come to terms with their sibling's learning disabilities (Gosling & Herbert, 1996). This essay aims to examine some of the factors which may impact on the relationships between siblings and their brothers or sisters with learning disabilities. Positive and negative implications of being the sibling of a child with learning disabilities will be discussed. The clinical implications of recent research findings will be highlighted. However, these discussions need to be placed in the context of past research, and most importantly, a discussion of the limitations of research in this field to date.

Methodological limitations of previous research

Unidirectional

Research on the sibling relationships of children without learning disabilities focuses on the bi-directional nature of relationships, and examines the effects that one child can have on the other and vice-versa. This is in stark contrast to the research on siblings of children with learning disabilities (Senapati & Hayes, 1988). Here, the majority of research concentrates on the effects of children with learning disabilities on their

siblings. Where the impact of the siblings on the child with learning disabilities is considered it is within the context of the non-disabled sib providing training or teaching to them.

Focus on negative effects

This unidirectional approach to assessing the impact of a child with learning disabilities on family functioning can lead to families which contain a member with learning disabilities becoming pathologised (Simeonsson & McHale, 1981). In fact, learning disabilities are common (Turk, 1996). By definition, 2 to 3 per cent of the population have an intelligence quotient below 70. Therefore it is not unusual for families to contain a member with learning disabilities, yet little research is carried out into normality of functioning for such families. Instead, research findings have concentrated on possible negative effects on siblings of children with learning disabilities. Increased child-care responsibilities (Blackard & Barsh, 1982), unreasonably high parental expectations of non-learning disabled sibs (Cleveland & Millar, 1977) and anger and resentment towards the child with learning disabilities (Wasserman, 1983) have been identified as possible detrimental effects on siblings of children with learning disabilities.

No, or inappropriate, comparison groups used

Findings of detrimental effects on siblings may be reliable, but without carefully controlled studies, the processes by which these detrimental effects manifest themselves cannot be examined. The majority of studies examining the effect of a child with learning disabilities on family functioning have not used any comparison groups (Howlin, 1988). This can result in an exaggeration of that child's role in causing problems for the family, where the reality is that sibling relationships where neither child has a learning disability are not universally harmonious.

Instead, it can be hypothesised that the negative effects on siblings arise from a number of factors, which are not unique to families containing a member with learning

disabilities. The environment of such families may be similar in important ways to other family situations. For example, one effect of being a sibling of a child with learning disabilities might be that parents have less time to spend on them. This could be similar to a child in a family where both parents work. Therefore, research on families with a child with learning disabilities should either employ carefully chosen comparison groups to examine the specificity of an effect, or conceptualise the research as a natural experiment in which the family provides an analogous environment to other family situations. Hence, examining the relationships between siblings and children with learning disabilities may give insight into mediating factors affecting other families (McHale & Gamble, 1989).

Inappropriate methods used in research

Research into “normal” sibling relationships employs a variety of research methods, including assessing each child’s perspective of the relationship and observation of the siblings (Dunn & McGuire, 1992). In contrast, research on families containing a child with learning disabilities have focused on the non-disabled child’s perspective, and relied heavily on questionnaires and interviews to assess their perceptions and feelings about the relationship (Senapati & Hayes, 1988). This is in part due to the research focus on effects rather than processes. Other research methods employed have included retrospective accounts of growing up with a child with learning disabilities and used teacher or parental report in addition to siblings’ self-report.

Each of these methods contributes to a static and incomplete account of the relationship between children with learning disabilities and their siblings. Although there are concerns about the reliable and valid assessment of the feelings and perceptions of children with learning disabilities (King, Josephs, Gullone, Madden & Ollendick, 1994), given appropriate methods of assessment children with learning disabilities can make reliable respondents (Silon & Harter, 1985). There is preliminary evidence that adults with learning disabilities can use questionnaire and personal interview formats reliably (Dagnan & Ruddock, 1995). Hopefully, future research

will attempt to assess the views of children with learning disabilities as a matter of course.

Retrospective accounts often find that siblings report that they benefited from growing up with a brother or sister with learning disabilities. Positive effects include enhanced maturity, compassion or sensitivity (Cleveland & Millar, 1977). However, these self-reports may not be reliable as siblings may selectively recall positive aspects of their childhood, or be able to report on them with more maturity and reflectivity than when they were children (Lobato, 1983). Furthermore, the population sampled may not be representative of all siblings of children with learning disabilities as participation in such studies is voluntary. Therefore siblings with negative experiences may find the process of reporting such experiences too painful and may simply have declined to take part.

A number of studies have used the ratings and report of teachers to enhance the external validity of their findings. For example, Blackard & Barsh (1982) gave parents and teachers a forced choice questionnaire assessing the impact of children with learning disabilities on their siblings. They found that teachers tended to overestimate the negative effects and had lower expectations of the siblings' adjustment. Studies which sample other sources of information need to take into account the possible biases or misperceptions of informants.

Social and political context of research findings

All research findings should be placed within a political and social context, as findings are in part specific to the prevailing opinions and laws of the time. For example, in the USA, the Education for All Handicapped Children Act was passed in 1975 (Wilson, Blancher & Baker, 1989). This law provided a broad range of publicly funded services to families with members with learning disabilities, increased publicly available information about disabilities, focused on mainstreaming children with learning disabilities and introduced initiatives to combat prejudice against people with learning disabilities. Wilson *et al* hypothesise that this increase in openness and information

about learning disabilities may enhance acceptance and hence reduce any negative effects on all family members.

In the UK, the recent emphasis on caring for people with learning disabilities in community settings may have similar effects in increasing their visibility and acceptance. Certainly, sensitive and moving accounts of problems and challenges faced by families with children with learning disabilities are frequently found in the national press (see, for example, *The Independent*, 7/5/97, or *The Independent* on Sunday, 11/7/97).

Clinical services are also operating within a rapidly changing context where the biological causes of learning disabilities are increasingly being identified (Turk, 1996). Knowing a definite cause of a child's learning disabilities can impact on the whole family system, for example by facilitating the grief process (Turk, 1996). The identification of different syndromes which can cause learning disabilities has led to the foundation of number of independent "syndrome societies" (Turk, 1997) which may be invaluable in providing support and advice to families. Services are conceptualising the impact of children with learning disabilities on the whole family system (Turk, 1996) and are increasingly recognising that these families may be especially at risk for experiencing difficulties. Increasingly services are acting proactively to offer services to groups who are conceptualised as being at risk for developing later problems (Gosling & Herbert, 1996). Carefully controlled, longitudinal studies are needed to examine whether prompt diagnosis and early intervention result in more positive outcomes for these families.

Summary

Many research studies to date have relied on unidirectional accounts of the relationships between children with learning disabilities and their siblings. These have often focused on the negative impact which a child with learning disabilities can have on their siblings. Many studies in this area have not used comparison groups, thereby limiting the specificity of findings. Where studies have reported positive outcomes for

siblings there has been an over-reliance on retrospective accounts from unrepresentative samples, augmented by others' reports which may be prone to bias. There is, therefore, a need for research to examine the relationships between children with learning disabilities and their siblings using bi-directional models which assess both children's perceptions and feelings about the relationship and incorporate direct observations. Finally, research findings need to be placed in the social and political context in which they occur. Important mediating variables in families' experiences may be affected by laws and provisions for children with learning disabilities, and the ways in which services conceptualise and act on the perceived needs of these families.

Recent research findings on the relationships between children with learning disabilities and their siblings

With the above caveats in mind, the next section contains reviews of recent research findings into the relationships between children with learning disabilities and their siblings. Wilson *et al* (1989) interviewed 24 children, aged 9-13, with younger siblings with severe disabilities. They found that these older siblings had a high level of day-to-day involvement with their siblings, accompanied by feelings of responsibility for their welfare. Despite these potentially negative factors, the siblings reported pleasure in their ability to amuse and care for the child. The majority reported that they got on "very well" with their siblings. However, they were also able to acknowledge their sadness, anxiety and anger. There was no effect of age or sex on any of these findings.

Thus the siblings assessed in this study seem to have a balanced view of growing up with a child with learning disabilities. There are undoubted stresses and downsides, but these siblings were not universally negative about the experience. However, there are a number of limitations to this study. First, there is no comparison group. Many of the findings may not be specific to families containing children with learning disabilities. Secondly, the families selected to take part in the study were already part of a larger research protocol, and had already had two in-home and two phone interviews over a period of two years. This suggests a high level of organisation and

motivation on the part of the families, who may not, therefore, be a representative sample of families containing a child with learning disabilities. Finally, the families had a high degree of on-going contact with services, which may have facilitated their adjustment to the child with learning disabilities and hence fostered more positive sibling relationships.

Stoneman, Brody, Davis & Crapps (1988) investigated the potential negative effect on siblings caused by assuming more responsibilities for caring for the child with learning disabilities. They compared 16 older siblings of children with learning disabilities with 16 older siblings of children without learning disabilities on measures of the amount of time spent on household tasks and childcare; the amount of peer contact and activities of both siblings and children with learning disabilities and made direct observations of the siblings interacting.

With respect to household tasks and childcare there was a strong effect of sex, with older sisters having more responsibilities for household chores and brothers for outside tasks, regardless of the status of their younger siblings. Older sisters of children with learning disabilities spent significantly more time baby-sitting than any other group, but brothers of these children spent as much time baby-sitting as sisters of children without learning disabilities.

Older brothers of children with learning disabilities had more friends to visit than any other group, and also visited others most frequently. Children with learning disabilities of both sexes had restricted peer contacts compared to any other group. They had significantly fewer peers visit and went to others' houses less frequently than younger siblings without learning disabilities.

The relationship between childcare responsibilities (as assessed by their self-report) and the observed sibling interaction was examined. There were several findings specific to the group of children with learning disabilities. Regardless of gender, older siblings with more childcare responsibilities had less positive interactions with their siblings. In addition, increased time spent on household tasks was associated with a decrease in

sibling conflict. Finally an increase in time spent on familial responsibilities was associated with a decrease in peer contacts for siblings of children with learning disabilities.

Important demographic variables may mediate these findings. For both groups the amount of time spent on childcare responsibilities was negatively correlated with family income and parental education. In contrast, out of home activities were positively correlated with family income and parental education. In the group of children with learning disabilities only, the amount of time spent on household tasks was positively correlated with the sibling's age. Finally, in the this group, the lower the adaptive competence of the child with learning disabilities, the greater the older sibling's responsibilities for their direct care.

There are a number of useful conclusions which can be drawn from this study. First, it seems that older sisters take on a disproportionate number of extra responsibilities for child care in families with children with learning disabilities. However, older brothers also have greater responsibilities in these domains than in families with no children with learning disabilities. Despite these greater familial responsibilities, siblings of children with learning disabilities did not spend less time with their peers, either outside the home or in having their friends visit. The finding that more time spent in childcare is associated with a higher degree of sibling conflict in the learning disabled group suggests that contact time per se can increase animosity between siblings (supported by the finding that increase time spent on other chores not in direct contact with the child with learning disabilities was associated with less conflict). Alternatively, there may be some aspect specific to caretaking which promotes conflict between siblings. Finally, regardless of whether the family contains a child with learning disabilities, the amount of extra responsibilities put upon older siblings is a function of family income, as is the amount of time they spend in out of home activities.

Finally, McHale & Gamble (1989) assessed 32 older siblings of children with learning disabilities and 32 older siblings of children without learning disabilities. The child was interviewed to assess their self-esteem, depression, anxiety and perceptions of their

interactions with their younger sibling. Maternal reports of the family background, child with learning disabilities' behaviour and functioning, conduct problems and the interactions between siblings were obtained.

They found that siblings of children with learning disabilities engaged in caretaking of their younger sibling more frequently than the comparison group. Older sisters spent the most time in caregiving, with older brothers of children with learning disabilities spending as much time in caregiving as older sisters of children without learning disabilities. On several measures there were significant effects of sex, not group. Girls reported more frequent negative interactions with their siblings and girls reported more involvement in household tasks than boys.

In terms of the sibling relationship, both children and mothers evaluated the relationship positively. Where there were between group differences they favoured the learning disabilities group. For example, siblings of children with learning disabilities were rated more positively in terms of their warmth and physical aggression. In addition, they reported that they were happier with how they got along with their siblings than older siblings of children without learning disabilities.

Finally the scores of siblings of children with learning disabilities on measures of well-being were lower than children in the comparison group. They had significantly higher levels of depression and anxiety, and lower perceived social acceptance and perceived conduct. There was a group by sex interaction, such that older sisters children with of learning disabilities had lower self-esteem than any other group. However, all the scores for the siblings of children with learning disabilities were within the normal range for the questionnaires used. It is difficult to interpret high scores which are below the clinical cut-off. One valid interpretation is that these scores are developmentally normal for siblings of children with learning disabilities.

Summary

The above recent studies, using appropriate comparison groups, maternal reports and measures of sibling interaction are more methodologically sound than earlier studies. They reveal that many factors, such as the amount of out-of-home activities engaged in by siblings may be more influenced by family's income than the presence of a child with learning disabilities. However, there is converging evidence that, despite cultural changes, older sisters of children with learning disabilities may still undertake a disproportionately large amount of caregiving, and as a group have lower self-esteem than other older siblings. In addition, older siblings, regardless of sex, have higher levels of depression and anxiety than other older siblings.

Clinical implications

The final section will outline some of the implications for clinicians of the findings and issues discussed above. First, it may be appropriate for services to act proactively, and intervene in families with children with learning disabilities from the point at which their learning disabilities are diagnosed and assessed. However, such early intervention, if insensitively targeted, could promote the idea that families containing children with learning disabilities are pathological, and all in need of help. Such a model is unhelpful (Simeonsson & McHale, 1981). There is a role for services in providing accurate and sensitive advice about the cause and course of a child's learning disabilities, and helping all family members come to terms with the meaning of the learning disabilities for them (Turk, 1996).

Clinicians should help the family in taking account of their own needs and those of their other children. There is converging evidence of increased responsibilities placed on older children, especially older sisters, which is associated with lower family incomes. Increased childcare responsibilities in turn are associated with more negative interactions between siblings. Clinical services have a direct role in providing information about benefits available to families, and services which they can access to alleviate the high demands for childcare which may inappropriately be placed on siblings. The latter includes respite and day care (Howlin, 1988) and contacts with

groups such as contact-a-family and the various “syndrome societies” (Turk, 1997). These can be invaluable normalising environments for families and provide practical advice from families who have experienced similar difficulties.

There is one important area of future research which has not yet been systematically evaluated. There is increasing awareness of genetic and chromosomal causes of learning disabilities. In many cases there may be direct implications for other family members of having a child with a syndromal cause of his learning disabilities. For example, other children may also be affected by the same genetic abnormality, as is the case of girls who are carriers of the fragile X gene which may cause severe learning disabilities in their brothers. Female fragile X carriers can show a behavioural phenotype which includes social anxiety, shyness and anxiety (Lachiewicz, 1992). If clinicians are unaware of the possibility of siblings’ difficulties being partially genetically determined this may lead to an incorrect assumption that the problems are caused by the presence of the child with learning disabilities.

Conclusion

Growing up in a family with a child with learning disabilities presents a number of challenges to the family. Contrary to assumptions in past research, the challenges faced by these families may not be unique, but may have overlap with other family set-ups. However, this is not to deny the stresses which families with a member with learning disabilities experience. Recent research has shown that siblings of children with learning disabilities may indeed experience a number of negative outcomes, with a robust finding that older sisters of children with learning disabilities take on disproportionately large responsibilities for childcare. This group also has the lowest self-esteem of groups of siblings of children with or without learning disabilities. This has to be counterbalanced against the positive aspects of the experience of growing up as the sibling of a child with learning disabilities. On the whole, siblings and their mothers report that the siblings’ relationships with the child with learning disabilities are positive.

There is a role for clinicians in acting proactively to help families who have children with learning disabilities, although this approach should not equate to pathologising these families. Clinicians are well-placed to provide information about the cause and course of a child's learning disabilities, which may facilitate meaningful planning for the child and family's future. An important part of clinicians' work is providing information on services to offset the increased childcare responsibilities which may negatively impact on siblings. In the future, research into families with children with learning disabilities may become more specific, and examine the particular needs which may arise from different causes of learning disabilities.

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Summary of Clinical Chapter

This chapter contains details of the six clinical placements completed during the course. During the first two years, four core placements were completed. The order of these placements was: Adult Mental Health; People with Learning Disabilities; Child and Adolescent and Older Adults. During the final year two specialist placements were completed in the areas of Children with Developmental Disabilities and Neuropsychology. The placements ran concurrently with the academic and research components of the doctorate and were each of six months duration. Typically, three days a week were spent at each placement.

A frontsheet detailing each placement is given, followed by a copy of each placement contract between trainee and supervisor. Then, a one page summary of a clinical report written on placement is presented. One clinical report is submitted from each core placement, in addition to a neuropsychological report completed whilst on a specialist placement. The clinical reports are submitted in full and in confidence in the second volume of this portfolio.

The reports selected for submission are a subset of those completed on placement. They have been chosen to demonstrate a range of client groups, problems and models used during the clinical component of this course.

Adult Mental Health Core Placement

Supervisor: Dr. David Brock

Location: Beechcroft Resource Centre

120 Victoria Rd

Horley

Surrey

Dates: October 1994-May 1995

DB/12/944

ADULT MENTAL HEALTH CORE PLACEMENT - CLAIRE GARNER

2nd November 1994 - 5th May 1995

Supervisor : David Brock

Aims of Placement

To provide experience of the full range of clients referred to adult settings; to expose trainees to a wide variety of service settings; to learn a specifically psychological approach to formulating problems; and to provide appropriate interventions.

Main Objectives (see Adult Placement Core Experience Document)

1. Exposure to a range of experience in a variety of settings.

Methods

Visits to a variety of wards and departments in the unit.

Visits to community facilities for mental health.

Regular attendance at a mental health resource centre including clinical liaison with staff.

Regular outpatient clinics, taking on patients referred for therapy under supervision.

Guided reading and discussion with supervisor.

2. Exposure to work with individuals, families and groups.

Methods

Seeing individual clients for assessment, therapy and vocational counselling.

Observation and use of a range of assessment techniques - formal and informal - including administration of WAIS-R.

Observation of a member of the department specialising in family/child work.

Guided reading and discussion with supervisor.

3. Exposure to work of other professionals groups and MDT's.

Methods

Clinical liaison with other professionals involved in specific cases.

Joint work with other professionals on specific cases and groups.

Observation of the working practice of other professionals.

Guided reading and discussion with supervisor.

4. Exposure to work with patients of a broad age range.

Methods

Taking referrals for assessment and therapy across the complete adult range, including at least one elderly case.

Visits to wards and day care facilities for the elderly.

Co-leading a problem-solving group for elderly clients in a day hospital.

Guided reading and discussion with supervisor.

5. Exposure to patients with long term disabilities.

Methods

Clinical practice including assessment and design of treatment programmes for individual chronic hospitalised patients.

Exposure to and involvement in the work of the member of the department specialising in rehabilitation.

Visits to inpatient and community facilities for the long term psychiatrically disabled.

Provision of vocational counselling for individuals suffering long-term psychiatric disability.

Guided reading and discussion with supervisor.

6. Understanding of the referral process and appropriateness of referrals.

Methods

Exposure to departmental policies and performance standards on the handling of referrals.

Discussion with supervisor on appropriateness and response to individual referrals.

Discussion with supervisor regarding selection, composition and setting up of groups.

7. Exposure to developing service provision.

Methods

Observing the continuing developments of service provision within the Trust.

Involvement in the evaluation of new planned services.

Secondary Objectives

1. Experience of teaching.

Methods

Attendance at the department's multi-disciplinary teaching events.

Exposure via reading and discussion to the work of the department's teaching specialist.

Involvement in other teaching events as negotiated with supervisors.

Accommodation comprises

- 1) Use of individual office, with phone, for regular client appointments.
- 2) Your secretarial support from identified secretary.

Supervision comprises:-

1. A weekly supervision session of 1 - 1½ hours with David Brock.
2. Observation by the trainee of supervisor providing therapy to patients in outpatient clinic.
3. Observation by supervisor of trainee providing therapy.
4. Exposure to developing service provision.

The supervision sessions cover the experience gained by the trainee under the above objectives; and in particular, focus on discussing clinical work (including the contents of tape recordings of individual therapy sessions) in relation to assessment, treatment and report writing.

Other Experiences

Attendance at departmental, DPAC and SIG meetings.

Attendance at psychological and multi-disciplinary training events.

Awareness via reading and discussion with supervisors of national and local policies and issues relevant to the profession and its development.

8th February 1995

Adult Mental Health Case Report: Summary

This clinical report details the assessment and intervention with Mr. Brown, a 45 year old practice manager, who had been referred to the Adult Mental Health Team by his G.P. He was experiencing anxiety and depression, which were beginning to interfere with his performance at work. In addition, he reported feeling paranoid about other staff members' attitudes towards him and found that his work-related concerns were impinging on his home and family life. Initial questionnaire assessments indicated that he was experiencing anxiety and depression of case level severity.

A detailed history was taken of Mr. Brown's home and work history. He had recently experienced changes in his role within his family, including losing both his mother and step-mother and becoming a grandfather for the first time. He had also been dismissed from a previous post as a practice manager.

His current difficulties were conceptualised in a cognitive-behavioural formulation, in which his unexpected dismissal had conflicted with a core belief about his own efficacy as a provider for his family and challenged his belief that he had to rely on his own abilities to solve problems. These had set up vicious circles which maintained the anxiety he was experiencing.

The intervention began with sharing the formulation with Mr. Brown, and actively involving him in the process of therapy. Tasks were set to complete in between sessions to prioritise his current difficulties and analyse the resources available to him to deal with them. During later sessions, the focus shifted from active problem-solving to identifying strategies to prevent relapse. The assessment questionnaires were readministered and showed that both his anxiety and depression scores had reduced.

Thus, Mr. Brown gained considerable benefit from a brief psychological intervention. The explicitly collaborative nature of cognitive behavioural work is hypothesised to have facilitated his engagement with the therapeutic process.

People with Learning Disabilities Core Placement

Supervisor: Mrs. Gill Koheeeallee

**Location: Sutton Community Team for People
with Learning Disabilities**

Farm House

Orchard Hill

Carshalton

Dates: May 1995-November 1995

MERTON AND SUTTON HEALTH AUTHORITY PSYCHOLOGY SERVICE

LEARNING DISABILITIES PLACEMENT PLAN

AIMS:

1. To provide experience of the main elements of a psychology service for people with a learning disability.
2. To provide some knowledge of the less common elements.
3. To familiarise with philosophies, policies and services for people with a learning disability at national and local level.

PHILOSOPHY

Date Achieved

1. Learn about normalisation philosophy.
2. Treat clients appropriately for their age.
- ~~3. Attend a resettlement meeting.~~
4. Discuss deinstitutionalisation.
5. Visit a residence in an institution setting.
6. Visit a residence in a community setting.
7. Use acceptable language in report writing.

COMMUNICATION

Date Achieved

1. Undertake an initial assessment interview with:
 - (a) a person with mild learning disability.
 - (b) a parent/relative.
 - (c) a paid carer.
 - (d) another professional.
2. Converse with a person with moderate learning disability.
3. Interact with a person with:
 - (a) profound learning disability.
 - (b) multiple handicap.
- ~~4. Learn Makaton Stage I and II.~~
5. Feed back to:
 - (a) a person with mild learning disability.
 - (b) a parent/relative.
 - (c) a paid carer.
 - (d) another professional.
6. Discuss communication issues in the learning disabilities field.

1
Contact S. Pinner

ASSESSMENT PROCEDURES

Date Achieved

1. Identify questions that can be answered by psychological assessments.
2. Apply and interpret:
WAIS-R
LEITER INTERNATIONAL PERFORMANCE
3. Apply and interpret a language assessment,
e.g. Communication Assessment.
Profile (CASP).
British Picture Vocabulary Scale (BPVS).
4. ~~Apply and interpret an educational assessment,~~
e.g. Neale Analysis of Reading Ability.
Schonell Tests of Reading and Spelling.
5. ~~Select an appropriate norm-referenced test for a client from the range available.~~
6. Apply and interpret at least 2 of the following:
HAMPSHIRE ASSESSMENT FOR LIVING WITH OTHERS (HALO).
✓ BEREWEEKE SKILL TEACHING SYSTEM.
SCALE FOR ASSESSING COPING SKILLS.
VINELAND ADAPTIVE BEHAVIOUR SCALES.
FUNCTIONAL PERFORMANCE RECORD.
STAR PROFILE.
7. Select an appropriate criterion referenced test for a client from the range available.
8. Undertake a functional analysis of challenging behaviour.
9. Discuss range of other assessments available.
10. ~~Assess the personal and social impact of chronic disability.~~
11. Discuss: Have a knowledge of:
(a) mental health.
(b) social impairment.
(c) genetic syndromes.

ELS1

INTERVENTIONS

Date Achieved

Carry out a behavioural intervention.

- (a) Consult the literature.
- (b) Do a behavioural assessment.
- (c) Collect baseline data.
- (d) Design programme.
- (e) Explain and negotiate the implementation of programme with residential/day care staff.
- (f) Monitor and evaluate programme.
- (g) Discuss maintenance of programme.
- (h) Select and apply, or be familiar with:
 - (a) social skills.
 - (b) advocacy.
 - (c) interpersonal relationships.
 - (d) self awareness.
 - (e) loss and bereavement.
 - (f) anger management.
 - (g) assertiveness.
 - (h) cognitive methods.
 - (i) relaxation.
 - (j) desensititation.

WORK WITH STAFF/OTHER PROFESSIONALS

Date Achieved

1. Identify the network of specialist and generic services and access these according to the needs of a client.
e.g. residential placement.
day placement.
social work.
community nursing.
psychiatry.
general practitioners.
occupational therapy.
physiotherapy.
speech therapy.
disablement resettlement office.
careers.
voluntary agencies.
education.
register manager.
2. Explain and, where possible, perform the generic keyworking role for a client within a multidisciplinary team.
3. Give a clear presentation of a psychological assessment/intervention within a multidisciplinary meeting.
4. Prepare, deliver and evaluate a presentation on a specific psychological topic to a group of staff.
5. Attend and critically evaluate:
 - a) community team meeting.
 - b) client review.
 - c) policy development meeting.
 - d) management meeting.

PROFESSIONAL DEVELOPMENT

Date Achieved

1. To write a clinical assessment report, to include method, outcome and interpretation.
2. To write an intervention report, to include formulation, method and outcome.
3. Chair a department meeting.
4. Write notes of a department meeting.
5. Do a journal club presentation to the department.
6. Discuss departmental goals and practices.
7. Have an understanding of the effect of current legislation on the service.
8. Attend weekly supervision.
9. Observe supervisor at work.
0. Be observed by supervisor at work.
1. Attend a Regional Special Interest Group meeting.

Signed: *Ch. Casner* 18/5/95 (name of Trainee)

Signed: *G. M. Kohecallee* 18/5/95. (G. Kohecallee, Supervisor)

People with Learning Disabilities Case Report: Summary

This report concerns the difficulties encountered by a woman with learning disabilities when moving from a relatively sheltered placement to a less containing environment. Maria was a 31 year old women who had recently moved into a community home for people with learning disabilities. She had been referred by the home manager who complained that she was inappropriately touching staff members.

The initial assessment comprised information from the home manager, other staff members and Maria herself. Prior to the community placement Maria had lived in a convent, where she had lived and been educated since childhood. She had a number of friends there, including a girlfriend with whom she had a sexual relationship. The initial formulation considered the inappropriate touching to arise from a number of factors including: her inadequate previous sex education; the lack of contact with her girlfriend and the inability of staff to maintain consistent boundaries of appropriate behaviour.

The intervention comprised elements to tackle each of these areas. She was referred to a women's group which provided education in sexual well-being and health. Regular visits to her former home were established. The staff group were given advice on appropriate boundaries for her behaviour.

The intervention was reviewed in the light of a disclosure made by Maria, further information becoming available about her family background and the escalating complaints about her inappropriately sexualised behaviour. As interventions based on the initial formulation had not been effective other formulations are considered, which draw on the importance of early bonding interactions in promoting later appropriate behaviour.

Child and Adolescent Core Placement

Supervisor: Mr. Nick Kerby-Turner

Location: Horsham Health Centre

Worthing Rd

Horsham

West Sussex

Dates: November 1995-April 1996

**This Contract is designed to set the parameters for CLARE GARNER in the
Child & Adolescent Psychology placement with
Nick Kirby-Turner in the Mid-Downs Health Authority**

INDUCTION PROCESSES

For Clare Garner to gain an understanding of the relationship of Child Psychology to services in Child Mental Health, and also in Child Health & Child Protection Services. Specifically:

- a) Observe Clinical Child Psychologists working in different settings
- b) Observe an Educational Psychologist at work
- c) Observe a Clinical Medical Officer conducting a developmental assessment
- d) Observe a Juvenile Court
- e) Attend a session in a playgroup
- f) Visit Colwood Adolescent Unit
- g) Visit Larchwood Children's Unit
- h) Visit the Family Therapy Clinic
- i) Become familiar with issues surrounding Child Protection Assessment
- j) Endeavour to observe children with Pervasive Developmental Delay

CLINICAL WORK

For Clare Garner to familiarise himself with the range of assessment procedures and therapeutic techniques by:

- a) Outpatient work in the Psychology Department. A variety of cases, reflecting the full age range, to illustrate the breadth of the speciality in terms of reasons for referral and therapeutic approaches applicable. Opportunities for individual and family centred work. Some joint work with Nick Kirby-Turner.
- b) Teaching:
Presentation in the Child Seminar Series to other Child Psychologists. Also, as opportunities arise, formal teaching of other professionals, possibly through case-based teaching.
- c) Research:
To discuss on-going research issues in child work.
To develop a small scale research project.

PROFESSIONAL DEVELOPMENT

For Clare Garner to endeavour to gain a perspective of service delivery issues through clinical work and to explore issues of service development by some attendance at Departmental Meetings and discussing issues as they arise.

SUPERVISION

At least 1.5 hours per week. Further supervision through informal meetings and via weekly Child Seminars. Some direct observation of Daryl Harris' work through joint session and the use of the VCR.

Child and Adolescent Case Report: Summary

This case report details the assessment and intervention offered to a family seen during the Core Child and Adolescent Placement. John, a seven year old, and his family had been referred to the Child and Adolescent Service after his mother had found deep, self-inflicted, scratch marks on his legs. He also suffered nocturnal enuresis and had difficulty sleeping. Self-injurious behaviour and enuresis are both uncommon in children of this age.

The initial formulation was based on two assessment interviews which John and his parents attended. An assessment of the presented symptoms aimed to identify the function of each behaviour for John. The assessment was broadened to include a systemic perspective on the family's current problems, which identified differences in parental style towards John, and clarified each family member's beliefs about the cause and nature of the current problems.

The initial formulation considered the function of nocturnal enuresis and scratching to be a dysfunctional way of seeking help, and sought to conceptualise this within a structural model of family dynamics. The intervention which was based on the initial formulation focused on teaching behavioural strategies to help John gain bladder control, and clarifying the boundaries within the family system. As the work progressed, the link between bouts of nocturnal enuresis and John's unresolved worries became clear: the intervention was modified to explore this issue.

The efficacy of the intervention was monitored by the number of consecutive dry nights achieved by John. By the end of the intervention he had achieved 12 consecutive dry nights, which represented substantial improvement. The ability of the initial formulation to encompass each family member's belief about the nature of the problem is discussed. Finally, the practical difficulty of working within a systemic model when only a subsystem was engaged in therapy is recognised and discussed.

Older Adults Core Placement

Supervisor: Mrs. Sue Webb

Location: 1 Oakhill

Surbiton Hospital

Surbiton

Surrey

Dates: April 1996-October 1996

KINGSTON & DISTRICT COMMUNITY NHS TRUST
Department of Clinical Psychology

CORE PLACEMENT - SERVICES TO OLDER ADULTS

CONTRACT FOR CLAIRE GARNER

AIMS

- To gain an overview of the services available for older adults in Kingston and District and an understanding of how these fit together to form an organisation.
- To gain an understanding of the role of the Clinical Psychologist providing services for older adults, and how this differs from other professions.
- to have experience of working in a variety of work settings, with a range of client groups and problems, and to develop skills and approaches when carrying out interventions, e.g. networking, liaising with other professionals, family therapy, marital work, behaviour modification, personal construct theory, bereavement counselling etc.
- To undertake some area of service development/project work/training.

OBJECTIVES

1.a To visit and meet staff in the following settings:

- Oak Day Hospital
- Wards for older adults with mental health problems at Tolworth Acacia Unit
- Wards for older adults with physical health problems at Tolworth Hospital
- Social services residential homes, e.g. Newent House, Murray House
- Private and voluntary residential homes
- Resource centres

1.b To spend time with a consultant psychiatrist, social worker and CPN, both in client meetings and, if possible, in individual client work.

2. To work with outpatients/community clients with the following problems:

- Anxiety
- Depression
- Phobias
- Bereavement issues including adjustment to disability
- Carers' issues

Core Placement - Services to Older Adults: Contract for Claire Garner (continued)

3. To observe and carry out neuro-psychological assessments using the WAIS-R, WMS or Coughlan, MEAMS or Mini Mental State.
4. To consult with staff in relation to problem behaviours in a residential setting.
5. To run a therapeutic group for older adults.
6. To have experience of a relatives support group.
7. To attend regular meetings held in the Psychology Department and other meetings within Services for Older Adults.
8. To have two hours supervision on a weekly basis.
9. To have one session study time per week.
10. To meet managers within the EMI Services and Elderly Community Services.
11. To carry out some teaching to staff and/or other colleagues.
12. To undertake an area of project work.
13. To gain experience of other areas of psychology work, such as health psychology and neuropsychology.

PERSONAL GOALS FOR CLAIRE

1. To have some experience working systemically, e.g. with families.
2. To carry out a more detailed neuropsychological assessment using a range of tests.
3. To have more assessment and rehab experience of working with a client suffering from a stroke, e.g. with Anna on the Stroke Unit.
4. To gain more experiences of working with clients suffering from behaviour problems.

SUE WEBB
Chartered Clinical Psychologist

CLAIRE GARNER
Psychologist in Clinical Training

Older Adults Case Report: Summary

This case report examines some of the issues facing families when one member is diagnosed with a neurodegenerative disease. Mrs. Barker was an 82 year old lady who lived with her husband and youngest daughter. She had been diagnosed as having Parkinson's disease. The referrer reported that the physical symptoms of Parkinson's disease were being exacerbated by her anxiety, and that this was also causing difficulties in the relationship between Mr. and Mrs. Barker.

The initial assessment comprised information from Mrs. Barker's medical notes and her personal and family history. Mr. and Mrs. Baker had long-standing marital difficulties, and acknowledged that they approached problems in different ways. The difficulties faced by Mrs. Barker and her family were formulated in terms of her reaction to having Parkinson's disease; how her behaviour and the advice and behaviour of others turned her impairment into disability and how the family system had reacted and adapted to changes in her physical health.

The formulation led to intervention on a number of levels. Mrs. Barker was offered individual sessions to explore her reaction to Parkinson's disease. The family were invited to contact the Parkinson's Disease Society to increase their knowledge of Parkinson's Disease and explore possible daytime activities for Mrs. Barker. Finally, Mr. and Mrs. Barker were seen together to explore ways in which Mrs. Barker could appropriately reclaim aspects of her past roles within the family.

The efficacy of the intervention is discussed with reference to the three strands of the intervention. The most successful aspect was the provision of individual support for Mrs. Barker. Limited progress was also made in other areas. The ongoing need for further intervention is noted.

Children and Adolescents with Developmental Disabilities Specialist Placement

Supervisor: Dr. Maria Callias

Location: Department of Child Psychiatry and

Clinical Psychology

St. George's Hospital

Blackshaw Rd

Tooting

London

Dates: October 1996-April 1997

CHILD SPECIALIST PLACEMENT

BETWEEN DR MARIA COLLINS, CLINICAL PSYCHOLOGIST SUPERVISOR

AND

CLAIRE GARNER, CLINICAL PSYCHOLOGIST IN TRAINING

Start Date:

Days Of Week: Wednesday, Thursday and Friday

To Include: **Assessment**

To gain experience of a wide range of psychometric assessments (developmental, neuropsychological and behavioural).

Number of assessments: Minimum of 6.

Therapy

To focus on one therapeutic model, but to include discussion of others as and where appropriate.

To gain experience of working systemically with families.

Number of treatment cases: Minimum of 4.

Experience

Experience of working with individuals and ideally group work with children with learning disabilities (over the complete age range 0 - 18).

To have the opportunity to observe the supervisor and to be observed.

To gain experience of a wide range of agency settings.

Aims

To gain experience of this client group.

To focus on refining assessment, intervention, therapeutic techniques and strategies.

Integrating formulation and treatment plans.

To start a final year research project.

To undertake a piece of research to satisfy the research on placement criteria.

To develop confidence in therapeutic style.

To consolidate working within a cognitive behavioural and behavioural framework.

Ch Garner

22/11/96

Dr Maria Collins

22 November 1996.

Children and Adolescents with Developmental Disabilities Case Report

(Neuropsychological Case Report): Summary

This case report details the assessment and intervention offered to a family whose son had suffered a severe head injury. Jerome and his family were referred to the Children and Adolescents with Developmental Disabilities Team for advice on how to manage the cognitive and emotional sequelae of a severe head injury which Jerome had suffered six years previously.

A complete developmental history showed that Jerome had been developing normally prior to the head injury. However, since the injury, and prior to the team's involvement he had suffered multiple further neurological insults including infections, epilepsy and hydrocephalus. A full cognitive assessment was therefore carried out in order to assess his current level of functioning, and to consider whether there had been any decline in his cognitive abilities since the accident.

The assessment revealed widespread and severe cognitive deficits, with particular impairments in everyday memory. The cognitive assessment was used to advise him, relevant professionals at school and his family on appropriate ways to compensate for his cognitive difficulties. This included advice on the most appropriate way to make use of his remaining cognitive abilities as well as the use of aids (such as diaries) to compensate for his impaired memory.

The initial assessment had also identified prolonged and unresolved grief in the family about Jerome's head injury and its sequelae, which was addressed in separate family work. The case report concludes with a résumé of the strengths and weaknesses of the case, including a discussion of the importance of individual work with the head injured child to assess their perception and beliefs about their impairment.

Neuropsychology Specialist Placement

Supervisor: Mr. Drew Alcott

Location: Unsted Park Rehabilitation Hospital

Munsted Heath

Godalming

Guildford

Dates: May 1996-September 1997

UNSTED PARK
HOSPITAL

NEUROPSYCHOLOGY PLACEMENT CONTRACT

FOR: Ms Claire Garner

DATES: May-September 1997

CLINICAL ACTIVITY

1. Follow 1 case through the initial interview, assessment, interpretation and formulation including drafting a letter/report.
2. To observe the administration of the following tests; BADS, AMIPB, Token Test.
3. To administer the AMIPB and BADS.
4. To see a variety of patients including those; minimally responsive state, cortical blindness, brain stem injury.

GROUP WORK

1. To feel moderately competent in conducting the 3 components of the Morning Orientation Meeting.
2. To attend the Brain Injury Group, to increase knowledge of brain injury.
3. To observe a variety of other groups including Review & Planning, Meal Preparation, Cognitive Group.

TEACHING SKILLS

1. To present a topic to the staff group.
2. To participate in the preparation of non clinical psychologists in test administration.

Cont./....

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PROPSYCHOLOGY PLACEMENT CONTRACT

ORGANISATION WORK

To discuss organisational issues with the supervisor.

SEARCH

To develop 1 idea from clinical practice which might potentially be progressed into a research. To develop this idea to the stage of drafting a research design.

SITS & MEETINGS

To attend the HIRU Clinical Review.

To attend 4 ward rounds in the General Neuro Team.

To attend a SIG meeting at Ticehurst.

To attend 2 team meetings in the General Neuro Team.

To attend 2 case conferences or family meetings (HIRU).

To visit Putney and Wolfson Units.

COURSES/TRAINING

No specific plans.

OTHER ACTIVITIES

To observe a variety of therapists at practice including; physiotherapy, occupational therapy, speech and language therapy and rehabilitation assistant's session.

me: Drew Alcott
Consultant Clinical Psychologist

Signed:

Drew Alcott

Name: CLAIRE GARNER
CLINICAL PSYCHOLOGIST IN
TRAINING.

Signed:

Ch Garner

te:

June 30, 1997

Date: 17.7.97

Summary of research chapter

The research chapter contains four separate pieces of research, completed throughout the course. The first is a literature review of issues relating to the assessment and classification of mental illness in people with learning disabilities. It reviews the methodologies which have been used to assess the incidence and prevalence of mental illness, and examines the validity of using existing classification schemes with this population. The review ends with a summary of current assessment tools used to assess mental illness in this population.

The small scale research project is an investigation of the ability of children with mild learning disabilities to use Likert and analogue response formats reliably. Reliability of self-report in people with learning disabilities generally has been relatively under-researched, and fewer studies have examined these issues in children. The results suggest that children with mild learning disabilities can make reliable respondents if materials are appropriately modified.

The research on placement is a brief report of a reminiscence group run for cognitively impaired older adults. The appropriateness of using groupwork in this population is examined, and strategies for successful groupwork discussed.

Finally, the large scale research project is a two group comparison study of the theory of mind and executive function performance of boys with fragile X syndrome and learning disabilities of unknown aetiology. The study of behavioural phenotypes, developmental neuropsychology and frontal lobe tasks is reviewed to give a complete rationale for the study. No group differences are found between any of the tasks used: this result is discussed in relation to the suitability of the tasks for the population studied.

Literature Review

The mental health needs of people

with learning disabilities:

Current Issues in Classification and Recognition

Assessment A15

Introduction

The mental health needs of people with learning disabilities are acknowledged to be underserved and poorly understood (Reiss, 1990; Lindsay, 1995). However, increasingly there is recognition that people with learning disabilities have a greater risk of developing emotional disorders than the general population (Sturmey & Sevin, 1993) and thus there is a growing need to investigate these disorders in people with learning disabilities. The difficulties of reliably and validly assessing the mental health needs of this client group will be discussed with reference to: the diversity of people with learning disabilities; the applicability of current classification systems and specific problems associated with eliciting and measuring psychopathology .

The exact prevalence of behavioural disorders and mental illness in this population are difficult to establish due to a number of methodological problems, including:

- the population studied (e.g. those known to services only or all people with learning disabilities in a given area);
- the definition of “mental illness” used (e.g. to include all behavioural problems or not);
- the definition of “case” used (e.g. those who already have a psychiatric diagnosis; those who are referred by carers or those who are identified using a specially designed screening or assessment tool).

However, there is general agreement that people with learning disabilities show a higher rate of psychiatric and behavioural problems than the general population. In a review of this area, Singh, Sood, Sonnenklar & Ellis (1991) conclude that:

1. about 8% to 10% of individuals with mental retardation in institutions have severe mental illnesses requiring treatment;

2. about 50% of people with mental retardation in institutions have at least one identifiable psychiatric disorder;
3. about 10% of institutionalised adults with mental retardation have severe mental illness, and about 60% have at least one severe or minor mental illness;
4. about 20% to 30% of institutionalised children with mental retardation have a mental illness, compared to 14% to 17% of the general population;
5. up to 30% of children with mental retardation living in the community have a diagnosable mental illness.

As Sood *et al* fail to specify which criteria they use to define “mental illness” and “psychiatric disorder” in this population, it is unclear if behavioural problems are being included in the above figures as mental illness. However, the conclusion is that people with learning disabilities show a range of emotional disorders, and have higher prevalence rates than the general population. One of the “Health of the Nation” document’s aims is to reduce mental illness in the learning disabled population (Marsden, Perry & Roy, 1995), therefore there is a pressing need for professionals who work with people with learning disabilities to be able to recognise and appropriately assess emotional problems in this client group.

Throughout this review, “learning disabilities” will be used in preference to “mental retardation” when referring to those individuals classified as mentally retarded according to DSM-IV (APA, 1994) criteria. This reflects current terminology used with this client group within the UK. However, when referring to older papers, “mental retardation” will be used if it has been chosen by those authors. People with learning disabilities who also have identified mental health needs are often labelled the “dually diagnosed”. This term has been criticised as being inappropriate (Singh *et al*, 1991) and unnecessarily stigmatising, as in other areas of mental health it is used to describe those with mental illness and substance abuse. Thus the term will be avoided, with “people with learning disabilities with mental health needs” being used in preference.

The heterogeneity of people with learning disabilities

People with learning disabilities are classified in DSM - IV by three criteria. These are:

- Significantly subaverage intellectual functioning (IQ < 70)
- Deficits or impairments in adaptive functioning such as communication, self-care, home living and social/interpersonal skills
- Onset before 18 years

Further, this group is subdivided into severity of learning disability (termed mental retardation in DSM - IV) along the following lines:

Mild mental retardation	IQ 50-55 to approx. 70
Moderate mental retardation	IQ 35-40 to 50-55
Severe mental retardation	IQ 20-25 to 35-40
Profound mental retardation	IQ level below 20-25.

The proportion of people in each category is very different. Scott (1994) reports that approximately 80% of the learning disabled population have mild, 12% moderate, 7% severe and less than 1% profound learning disabilities. Further, there are striking differences in the characteristics of different levels of learning disability. Those with mild learning disabilities (IQ range of approximately 50 - 70) typically show little impairment in social functioning. They are less likely to have an organic impairment or physical handicap, but more likely to have siblings and parents of lower than average IQ. In this group, presentation of psychopathology is likely to be similar to the general population (Scott, 1994; Fletcher, 1995). In contrast those moderate and more severe leaning disabilities often show marked impairment in social functioning. Parents and siblings tend to be of normal intelligence. This group also show a higher proportion of

genetic and chromosomal abnormalities such as fragile X syndrome (Scott, 1994). The presentation of psychopathology in this group may be very different to the wider population (Scott, 1994).

Thus within the client group of people with learning disabilities there are those who are at the low end of the normal distribution of IQ in the general population, but who have no specific biological cause of their learning disabilities, and those individuals whose learning disabilities are biological in origin (Murphy, 1994). For the purposes of this review it is more practical to consider people with learning disabilities in these two groups. Therefore, for the rest of this review, the group will be split into two along these lines, with mild learning disabilities used to indicate those individuals with no specific biological cause of their learning disability whose IQ is about 50 or above, whilst severe will refer to those whose learning disabilities have an identifiable organic aetiology, who tend to have a measured IQ below 50.

The interpretation of behaviours shown by people with learning disabilities

The differentiation of this client group is important when considering the interpretation and significance of behaviours in the learning disabled population. A key question in determining psychopathology in people with learning disabilities is the extent to which overt behaviours can be interpreted as indicative of psychiatric disorder. A classification system such as DSM-IV relies on measurement of observable behaviours to form the basis of diagnosis. Therefore, a large portion of this review will necessarily be concerned with the reliable and valid interpretation of observable behavioural signs.

There are a number of different ways of interpreting behaviours in this population, which make different assumptions about the factors which maintain such behaviours, and also logically lead to different intervention strategies. For example, one psychological model of an individual's behaviour would start from the assumption that the behaviour, however maladaptive or superficially random and unpredictable, would

serve an important function for that person. (LaVigna & Donnellan, 1986) and be reinforced (positively or negatively) by the environment they were in. A careful behavioural, functional analysis could then start to identify those variables which were maintaining the behaviour, and design a program which would aim to construct more adaptive ways for the person to attain the desired outcome. Recent advances in analogue techniques for identifying the maintaining variables of behaviour (Iwata, Dorsey, Silfer, Bauman & Richman, 1982); the development of reliable checklists for assessing motivational factors (Durand & Crimmins, 1988) and the expansion of the functional analysis to include a wider ecological perspective (Hastings & Remington, 1995) have provided a paradigm for understanding the processes which may underlie some behaviours in this population and given a rationale for developing appropriate interventions (Emerson, 1992).

A rather different psychological perspective is to use psychotherapeutic approaches to understand behavioural disorders in people with learning disabilities (Gaedt, 1995). Historically, people with learning disabilities were not thought to be suitable candidates for classical psychoanalysis as they lack the stable ego functions and reliable object constancy necessary to engage in the psychotherapeutic process. However, recent expansions in the basis of psychoanalytic theory has allowed explanations of the specific forms of psychopathology associated with learning disabilities. Gaedt describes:

- the transformation of organic brain deficits into mental disorders; in which the chief psychological sequelae of a learning disability is the disruption of early interactions, and its consequent impact on the development of psychic structures and functions;
- the secondary psychosocial deficit which develops as the interaction between infant and caregiver is of a different nature than the typical course of a child's development;
- the importance of the therapeutic relationship in recognising which developmental processes are being acted out by the client;

- the interpretation of repetitive and apparently maladaptive behaviours as transference phenomena, in which the client is affirming and experiencing their identity.

Whilst it is likely to be true that the early interactions of those with learning disabilities are different to the wider population, and that psychosocial deficits are a defining feature of people with learning disabilities, there are some difficulties in using Gaedt's model to understand the range of behaviours shown by people with learning disabilities. First, his description turns "learning disability" into a form of psychopathology itself, an approach which has been rejected from DSM-III-R (APA, 1987) onwards. Secondly, his assertion that behavioural techniques can only be effective on behaviours which have been "learned" is incorrect. Even if the precipitating factor(s) for a certain behaviour is unknown, behavioural techniques can be employed to modify the contingencies which currently maintain it (see Murphy, 1994). Finally he interprets behaviour therapy as a means to "eliminate" certain forms of behaviour. This is a crude interpretation of behavioural techniques, which can be used in a constructive way to allow a client to learn new, adaptive forms of self-expression.

Further, when considering those with severe learning disabilities it is recognised that certain syndromes have a specific behavioural phenotype which accompany them (Flint & Yule, 1994). For example, Cornelia de Lange syndrome is characterised by self-injurious behaviour (SIB), aggression, hyperactivity, hypersensitivity and autistic features (Tuinier & Verhoeven, 1993), whereas individuals with Prader-Willi syndrome invariably overeat (Murphy, 1994; Tuinier & Verhoeven, 1993). These authors warn against viewing all abnormal behaviours as being indicative of mental illness, as they may be syndrome-specific.

However, just because a behaviour is associated with a given biological disorder it is recognised that it may also be influenced by environmental and operant factors (Murphy, 1994). Thus, behaviours associated with a given syndrome can come to be controlled by environmental contingencies (LaVigna & Donnellan, 1986). Therefore, an integrated view of an individual's behaviour requires an appreciation of the combination of biological, environmental and operant factors which can contribute to the onset and maintenance of a given behaviour.

Classifying psychopathology in people with learning disabilities

Research into psychopathology in people with learning disabilities has been hampered by the lack of a valid and reliable classification system. The arguments commonly used in favour of classification in the wider population are very salient to promoting the mental health needs of people with learning disabilities (Matson & Frame, 1986; Sturmey & Sevin, 1993). First, classification facilitates communication between clinicians and researchers, and thus the development of models of different disorders. Secondly, classification aids the selection of the most appropriate intervention. Finally, classification aids research.

Although most researchers and clinicians agree that classification is important in this group, there are profound differences in which classification systems are seen to be appropriate, which behaviours should be included in a classification and on what dimension they should be classified. The next section will consider the various different ways in which classification can be considered, and following this measures which have been derived from these classifications reviewed.

Appropriate classification systems for use with this client group

Early papers concerning the prevalence of psychopathology in people with learning disabilities considered that standard classification systems could be applied wholesale

to this client group. Sovner & Hurley (1983), in a review of 25 published studies of affective illness in people with learning disabilities, reported “definite” diagnoses of depression according to DSM-III (APA, 1980) criteria in clients with mental retardation of mild to profound severity. They concluded that the presence of learning disabilities did not preclude the development of affective disorders, and that DSM-III criteria were appropriate to classify these disorders. However, they add the caveat that there may be differences in the way symptoms manifest themselves, especially as there may be communication impairments which makes accurate and meaningful identification of thoughts and feelings difficult. Thus this undermines the reliability of the diagnostic system, as researchers have had to interpret a behavioural sign to be equivalent to a given diagnostic criterion.

DSM-IV endorses the view that one set of criteria is appropriate for all client groups, giving a single set of criteria for most disorders which can be applied to children, adolescents and adults, although the presentation of a disorder may vary with the developmental stage of the individual. This is clearly a pertinent point when considered in relation to people with learning disabilities, whose developmental stage rather than chronological age may be a more appropriate framework within which to interpret behaviours (Dosen, 1993; Tuinier & Verhoeven, 1993).

However, even given this adaptation to standard DSM-IV use, the rationale of applying wholesale classification systems designed for a population of normal intelligence has been questioned. Campbell & Malone (1991) point out that diagnostic systems have several limitations when applied to people with learning disabilities. They can be difficult to apply, as they have not been designed to assess people with learning disabilities who have specific difficulties in reporting feelings and information. Further, the standard use of classification to aid intervention is difficult as people with learning disabilities are not usually included in comparative trials of treatments. Therefore these interventions are not validated with this client group. Thus, the difficulties in applying standard criteria to people with learning disabilities fall into two

categories: those concerned with the difficulty in eliciting information from the client (Sovner, 1986) and those addressing the validity of using these criteria within this population. The latter point will be discussed below, with the difficulties in use addressed in the final section.

Given the diversity of this population it is likely that existing classifications such as DSM-IV may be applicable for use with some of this client group. DSM-IV has been developed from studies on a population with a normal IQ distribution, with a mean IQ of 100 and a standard deviation of 15 (Campbell & Malone, 1991). One assumption, then, is that the criteria should be equally applicable to persons at all points in that normal distribution. Therefore, there is no *a priori* reason not to use DSM-IV criteria for people with mild learning disabilities, any more than there would be to classify psychopathology differently for those with IQ scores two standard deviations above the mean. Indeed to use a reliable, multi-axial system such as DSM-IV may counter some of the reported problems in assessing the mental health needs of this population such as diagnostic overshadowing (Reiss, Levitan & Szysko, 1982). This refers to the phenomena of the label of "learning disabilities" causing psychopathology to be overlooked. Spengler, Strohmer & Thompson Prout (1990) sent behavioural descriptions to mental health professionals who were asked to make a diagnosis. The descriptions of behaviour were identical, with the only difference being the IQ of the client. These were found a robust diagnostic overshadowing effect with people with an IQ of 58 less likely to be given a diagnosis of any mental disorder than those with IQ 70, 80 or 108. Professionals who had worked in the mental health field longest showed the most overshadowing.

Diagnostic criteria may be usefully and reliably applied to those with mild learning disabilities as many will be able to describe the same range of symptoms as the normal population e.g. express feelings of hopelessness, elation or sadness. Pawlarcyck & Beckwith (1987), in a review of symptoms of depression displayed by people with learning disabilities concluded that people with mild learning disabilities show similar

symptomatology to nondisabled people, whereas depressive symptomatology had to be inferred from behaviour in those with severe learning disability. Thus, in those with mild learning disabilities, a behavioural interpretation of emotional distress can be validated by self-report, as in the general population.

A further difficulty in applying diagnostic criteria to people with severe learning disabilities is the extent to which behaviours shown by this population can be interpreted as underlying signs of psychopathology. As mentioned above, people with learning disabilities often have impairments in communication and are therefore less able than the general population to report on their thoughts and feelings. Therefore, internal states are sometimes inferred from overt behaviours. This issue is very complex as there is an entirely unclear relationship between behavioural problems and mental illness in this population (Dosen, 1993; Holland & Murphy, 1990). Many studies chose to measure the prevalence of psychiatric problems and behavioural disorders together (Rutter, Tizard, Yule, Graham & Whitmore, 1976) which may be misleading in determining the mental health needs of this population. Murphy (1994) describes the interpretation of all behavioural problems in terms of psychiatric disorders as unreliable and often unhelpful. However, it is equally unlikely that behavioural problems never indicate underlying mental health needs. The conclusion reached by a number of researchers (Borthwick, 1988; Holland & Murphy, 1990; Dosen, 1993) is that there is an unclear area of overlap between behavioural disorders and underlying psychopathology.

Further, it is unclear which dimension of behaviour should be included in a classification system. The same topography of behaviour can be used by two different people to obtain different outcomes (e.g. head-banging gains attention for one client, but escape from task demands for another), indicating that classifying behaviours by outward appearance may not be useful. Therefore, other researchers have tried to derive empirical taxonomies of behavioural problems. Bruininks, Bradley & Morreau (1991) describe a taxonomy based on a hypothetical continuum of maladaptive

behaviour. This is subdivided into three descriptive categories of internalised, asocial or externalised behaviour, which in turn describe different types of problem behaviour. For example, behaviours which are hurtful to self, unusual or repetitive, or characterised by withdrawal are termed internalised. Others advocate that behaviour be classified according to its function (Tuinier & Verhoeven, 1993), an approach which has been adopted within the psychological literature (Holland & Murphy, 1990) and is formalised in the functional approach to analysing behavioural problems described above.

However, there is no clear way of mapping the empirical taxonomies of behaviour of the type given above onto existing systems which aim to classify psychopathology. Even if a behaviour can be controlled by manipulating consequences and antecedents this does not give a rationale for its appearance, nor exclude an underlying affective disorders as its cause. Some researchers use the idea of "behavioural equivalence" to determine which behaviours are characteristic of a disorder (Marsden *et al*, 1995). In their study they administered a checklist of psychopathology to carers of 82 residential clients with varying levels of learning disability, and also screened medical records, interviewed the client and monitored treatment response in order to investigate the clinical features of depression, according to ICD-10 criteria, in this population. They found that depression in moderately disabled clients was characterised by negative affect, isolation, self-injurious behaviour, reduced communication and weight loss, whilst in the severely and profoundly disabled group negative affect, screaming, aggression, sleep disturbance, self-injurious behaviour and reduced communication were common.

Unfortunately, however one chooses to map overt behaviours onto descriptions of symptoms there will always be a lack of independent standards against which to validate this interpretation. For example, there are few reliable biological markers of affective disorders, and those which exist have generally been developed on a non-learning disabled population. The dexamethasone suppression test (DST), widely

recommended as a aide to the detection of depression in those with severe learning disabilities (Pawlarczyk & Beckwith 1987; Singh *et al*, 1991), has been shown to give a high rate of false positive results (Mudford, Barrera, Murray, Boundy, Caldwell & Goldberg, 1995). Mudford *et al* (1995) tested 40 adults with severe learning disabilities, using three rating scales of behaviour and the DST. Nine participants gave true-positive results (i.e. the DST and checklist scores were in agreement) but seven were false positive. Moreover, those who gave a positive DST did not show a consistent behavioural profile, suggesting that a positive DST is not a valid measure of any given constellation of behaviours, let alone those which are believed to be the equivalents of behaviours typical of depression in the wider population.

Similarly, response to pharmacotherapy cannot be used to check the validity of a behavioural interpretation, especially in the severely learning disabled population. First, there is an inconsistent pattern of drug response in the wider population. Hollon, Shelton & Loosen (1991), in a review of pharmacotherapy treatments of depression, report that only around 50-60% of patients in drugs trials respond to medication. Thus, if the same drug is used to treat a client with learning disabilities and has no effect, the client may still be depressed but not pharmacologically responsive to that drug. In addition, it is not uncommon for people with severe learning disabilities to have idiosyncratic reactions to drug regimes, as many of the organic brain disorders associated with severe learning disabilities are characterised by abnormalities of neurotransmitter function (Tuinier & Verhoeven, 1993).

Even given these difficulties there are reasons for beginning an investigation into the presentation of psychopathology in people with severe learning disabilities by using diagnostic criteria:

- it provides a framework which will facilitate the development of reliable and valid assessment instruments

- its multi-axial structure should help counter diagnostic overshadowing
- it provides a basis for communication.

Instruments for eliciting psychopathology

In the past fifteen years there have been a number of attempts to standardise assessment of psychopathology in this population. Some of the difficulties in assessment have been outlined above, namely the use of classifications which have not been designed for the learning disabled; communication problems which hamper the accurate reporting of internal states and the differentiation of signs of psychopathology from other sorts of behaviour. To these, Sturmey, Reed & Corbett (1991) add: frequent use of third party reports of a client's behaviour, often without checks on their reliability and validity; the difficulties many clients have in maintaining records of thoughts and behaviours and the relationship of any diagnosis to a treatment plan. Three current instruments designed to assess psychopathology will be evaluated, with reference to the issues raised above.

Psychopathology Instrument for Mentally Retarded Adults (PIMRA)

The PIMRA (Matson, Kazdin, & Senatore, 1984) was developed around DSM-III criteria. It contains 56 items, on seven scales of: Schizophrenic Disorder, Affective disorder, Psychosexual disorder, Adjustment disorder, Anxiety disorder, Somatoform disorder, Personality disorder and Inappropriate Mental Adjustment. Self-report and informant versions were developed. The authors reported high test-retest reliability and internal consistency of the scales. However, concurrent validity was low across the two versions of the scale. Further, factor analytic studies showed little correspondence between the factor solution and the initial factors on which the instruments were based. The self-report version yielded two factors, labelled Anxiety and Social Adjustment, whilst the informant version gave Affective, Somatoform and Psychosis factors. Later independent attempts to investigate the psychometric

properties of the scale gave lower reliabilities and internal consistencies than those originally reported (Watson, Aman & Singh, 1988) and failed to recover the factors found by the first authors. These factors make the scale less robust than desirable. Further, the low concurrent validity suggests that the two versions of the scale may not be assessing the same factors.

The Diagnostic Assessment Schedule for the Severely Handicapped (DASH)

The DASH (Matson, Gardner, Coe & Sovner, 1991) is unusual in that it has been designed specifically for use with severely learning disabled clients, a group acknowledged to be particularly hard to assess (Sturmey *et al*, 1991). It is based around DSM-III-R criteria and previously published research, and takes the form of a checklist, designed to be administered to carers who are familiar with the client. The checklist is divided into 13 disorder groups, with items taken from DSM-III-R according to two principles: appropriateness for severe/profound learning disabled clients and comprehensibility to informants without formal training. The symptoms in each group are rated on frequency, severity and duration by the informant.

The internal consistency of the scales range from $\alpha=.82$ for elimination disorders (control of bowel and bladder) to $\alpha=0.20$ for schizophrenia. A recent study (Sevin, Matson, Williams & Kirkpatrick-Sanchez, 1995) reports high test-retest reliability across the scale items of .84 for frequency, .84 for duration and .91 for severity. Kappa values and intra-class agreements were consistently low (less than 0.4 and 0.5 respectively) for the anxiety disorders and schizophrenia subscales. The authors conclude that establishing the reliability of the scale is only beginning, and that any investigation of its validity and the relation to treatment is some way off.

The Psychiatric Assessment Schedule for Adults with a Developmental Disability (PAS-ADD)

The PAS-ADD has been developed from the Psychiatric Assessment Schedule (Dean, Surtees & Sashidharan, 1983), which itself was developed from the Present State Examination (PSE: Wing, Cooper & Sartorius. 1974). Thus it has the advantage of being based on an existing instrument which had reliability data associated with it for comparison with the developing schedule. It is a semi-structured interview which incorporates several features which make it easier to use with a learning disabled population:

- parallel informant and client questionnaires which can be combined to increase sensitivity;
- A three tiered structure, allowing a basic interview with a client with minimal verbal communication;
- the use of a memorable “anchor event” in the client’s life to help focus their attention on recent changes;
- items organised to make the interview procedure as simple as possible for the client to participate in;
- clear instructions to the interviewer as to which questions can be omitted, conditional on previous responses. This aims to keep the interview focused, and minimise the loss of attention by the client.

(Moss, Patel, Prosser, Goldberg, Simpson, Rowe & Lucchino (1993)).

Thus the interview is designed to allow the client to participate as much as possible, which provides information on the inter-rater reliability of the scale, and allows the client the basic dignity of reporting as far as possible on their current emotional state. The PAS-ADD aims to examine psychiatric symptoms only, and does not attempt to equate behaviours with symptoms. In the authors’ opinion, behavioural problems are best investigated by functional analysis coupled with informant interviewing. The

authors report the initial inter-rater reliability of the scale to be comparable to those obtained by Wing *et al* (1977), although the raters used in this pilot study were specially trained in using the PAS-ADD. However, this is a good basis on which to try the schedule more widely.

In a prevalence study using the PAS-ADD (Patel, Goldberg & Moss, 1993), the schedule was successfully used to interview a group of clients with an average IQ of 39. Of their sample of 105 clients, 65 were interviewed and able to provide some meaningful, clinical information whereas information on the remaining 40 clients was provided by informants only. They identified 12 cases: three from combined client and informant data, four from client data only and five from informant data only. These 12 cases were given some concurrent validity by an independent psychiatrist arriving at the same diagnoses. These results give rise to some important points:

1. when behavioural problems are excluded, the prevalence of mental illness estimated in this population is 11.4%. This is low when compared to the figures quoted by Singh *et al* (1991).
2. There is only 25% agreement in diagnosis between clients and informants who know the clients well. Thus any schedule which relies only on informant data is likely to give different results to one which combines client and informant responses.

Cases were diagnosed by informant only when the client was only able to give a partial interview. In those cases which the client only identified, the informant was either aware of the symptoms but ignorant of their clinical significance or had not noticed the symptom at all. Patel *et al* (1993) attribute this to either diagnostic overshadowing or a lack of clinical training.

The PAS-ADD encompasses many desirable aspects of a clinical schedule. First, it has been specially adapted for use with a learning disabled population, and makes

provision for the communication and cognitive difficulties likely to be encountered in this client group.⁴ Secondly, it has been developed from an existing, reliable schedule, allowing comparisons between current and previous research on reliabilities. It contains the original symptoms described in the schedule and does not try to substitute behavioural equivalents which may be invalid. Thirdly, the difficulties of using third party reports have been investigated, the potential discrepancy between reports highlighted, and reasons for this discrepancy given. Finally, the PAS-ADD has a defined function: to arrive at diagnoses. The PIMRA and DASH are essentially checklists which impose arbitrary cut-offs of symptom scores to indicate diagnoses.

The PAS-ADD is funded by the Department of Health, with the aim of producing a comprehensive ICD-10 clinical interview. Future developments are to include a brief checklist of symptoms to be used by direct care staff and a more detailed screening instrument (the mini-PAS-ADD) to be used by other clinicians (Moss, 1995). Despite all the positive points in its development, the PAS-ADD still takes as its basis a classification system which has not been validated on this population. Most researchers agree that it is likely that disorders and syndromes may present with a very different pattern of symptoms and time course, and that certain symptoms may not develop at all. However, there is considerable disagreement as to whether this should be resolved by investigating possible symptoms according to general classifications (the approach used in the PAS-ADD) or identify syndromes in a bottom-up fashion: by identifying symptoms and then factor analysing them to arrive at an empirically derived classification.

Conclusion

The learning disabled population is acknowledged to have serious and unmet mental health needs. Whilst those with mild learning disabilities may be able to benefit from the sensitive application of existing classification systems to investigate and inform their treatment, there is still much work to be done in identifying and recognising signs

of mental ill-health in the severely learning disabled population. This is complicated by a lack of external sources with which to validate research. The main problem would seem to be the difficulty in interpreting behavioural signs. Most behaviours are a combination of biological, operant and environmental factors, rather than either a substitution for communication or a psychiatric sign. Although work is beginning to be done into ways of reliably classifying disorders seen in this client group, it is clear that much empirical research is needed to investigate clusters of behaviours shown in this client group, and to identify appropriate and helpful dimensions along which they can be classified.

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Small Scale Research Project

**An initial investigation into the use of Likert and visual analogue
response formats by children with mild learning disabilities**

Assessment A37

Introduction

This study investigates how reliably children with mild learning disabilities can use visual analogue and Likert response formats. To date, there has not been a great deal of research into the reliable use of different response formats with people with learning disabilities, and fewer studies have examined these issues in children. Therefore, general issues pertaining to the importance of obtaining reliable self-reports from people with learning disabilities will be reviewed, followed by reviews of studies which specifically examine reliability of responding of both adults and children with learning disabilities.

Obtaining reliable self-report from people with learning disabilities

People with learning disabilities are vulnerable to a higher rate of affective disorder than the general population (Sturmey & Sevin, 1993), leading to a growing recognition of the need to identify and measure factors which may predispose this group to developing emotional disorders. Consequently, there has been a recent increase of research examining the reliable and valid measurement of thoughts and feelings of people with learning disabilities (Lindsay, Mitchee, Baty, Smith & Millar, 1994; Lindsay & Mitchee, 1988).

However, it is recognised that there are a number of factors which complicate the use of self-report measures with people with learning disabilities. First, most existing questionnaires measure concepts which have been developed from work with non-learning disabled populations. These may be neither reliable nor valid when working with clients with learning disabilities (Sturmey, Reed & Corbett, 1991). For example, using DSM-IV (APA: 1994) to classify disorders in people with learning disabilities may be inappropriate. This problem is likely to be more serious with more severely learning disabled populations due to: greater difficulties in verbal communication; the difficulty and validity of distinguishing functional and communicative behaviours from

psychiatric symptoms (Holland & Murphy, 1990) and the increasing proportion of people whose learning disabilities have genetic or chromosomal cause. The presentation of psychiatric disorders in these people is likely to be very different to those without an organic cause of their learning disabilities (Scott, 1994).

Secondly, there is a difficulty in finding independent measures against which to validate clients' self-report, once reliable methods of self-reporting have been established. The establishment of reliability has to precede investigations of validity. If existing classification schemes are inappropriate for this client group, there is no reason to expect that measures will have the same concurrent validity as in the group which was used to develop the measures. Some biological markers of disorders have also been shown to be unreliable in this group (Mudford, Barrera, Bellack & Senatore, 1995).

Thirdly, clients with learning disabilities may experience difficulties in understanding the structure of a scale or concepts examined. Self-report questionnaires are heavily dependent on receptive language competence (Reynolds & Baker, 1988). Lindsay & Mitchee (1988) report that clients may have particular problems in understanding the gradation of severity of a symptom, whereas Williams & Asher (1992) consider that children with learning disabilities may not be able to use formats where a 'true' response represents a negative state of affairs. Finally, people with learning disabilities may be particularly susceptible to a number of response biases such as acquiescence, faking good or bad responses or responding irrespective of the content of the question (Reynolds & Baker, 1988).

Reliability of self-report of adults with learning disabilities

In view of these difficulties, it is unsurprising that most research on self-report has been carried out with people with mild or moderate learning disabilities. According to Scott (1994) these are most likely to express disorders in the same manner as the

general population. This section will briefly review studies with adults and summarise the strategies used to enhance reliability.

The assessment of depression is a relatively widely researched area in adults with learning disabilities. For example, Helsel & Matson (1988) investigated the use of self-report measures of depression in 99 adults with borderline to severe learning disabilities. The self-report measures used were the Beck Depression Inventory revised for use with adults with learning disabilities (Kazdin, Matson & Senatore, 1983), the Zung Self-Rating Depression Scale - Revised for adults with learning disabilities (Kazdin *et al*, 1983), and the Social Performance Survey - Revised for Mentally Retarded Adults (Matson, Helsel, Bellack & Senator, 1983). The scales were scored using Likert scales, which were augmented with bar graphs to illustrate the concept of gradation. Scoring formats were demonstrated to each participant and responses were modelled. Understanding of the response format was assessed before each scale was administered, but no details were given of the criteria for considering a person to have understood the task demands. No reliability data were reported for any of these scales.

Reynolds & Baker (1988) developed the Self-Report Depression Questionnaire (SRDQ) specifically for use with people with learning disabilities. It is based on the symptoms of depression delineated in DSM-III (APA, 1980) and Research Diagnostic Criteria, worded in simple statement form and scored using a three-point response format. It includes reverse-keyed items to check that informants are not dissimulating. Their participants were 103 community-dwelling adults, with borderline to moderate learning disabilities (DSM-IV criteria). They administered a screening test (Social and Prevocational Information Battery - Form T: Irvin, Halpern & Reynolds, 1977) to ensure that their participants could answer the SRDQ without response bias. This battery consists of a number of minimal content questions (e.g. "The sun shines at night") which are answered yes/no. Participants who score below cut-off are considered to be unable to respond validly to the test battery.

The SRDQ had a high internal consistency (0.90 on first presentation, 0.93 on retest), and a test-retest reliability of 0.63 after 11 weeks. This represents moderate test-retest reliability, which is to be expected for a concept such as depression. Reynolds & Baker (1988) also investigated the criterion related validity of the SRDQ by correlating the results of the SRDQ with the Hamilton clinical interview. The correlation between measures was 0.63 ($p < 0.001$), supporting the validity of the SRDQ as a measure of depression in this population.

In contrast, Rojahn & Warren (1994) compared the performance of 38 adults with mild to moderate learning disabilities on the SRDQ, a standard psychiatric interview and an informant rating scale. They reported very low correlations between different measures and concluded that there was no meaningful relationship at all between them. Various explanations can be put forward for this lack of convergent validity, including lack of reliability in the psychiatric diagnosis, and the difficulties in informants interpreting others' behaviour as evidence of symptoms. However, it is curious that this study found almost no relationship between two self-report measures, which casts doubt on their criterion validity.

The results of Rojahn & Warren (1994) are somewhat anomalous. Lindsay *et al* (1994) examined the self-report of 67 participants with mild to moderate learning disabilities on a range of measures of depression and anxiety. They found a considerable degree of convergent validity in the results, with all measures of anxiety correlated together, and all measures of depression being correlated. No correlations were found between an extroversion scale and any of the measures. This would have been expected if the correlations were due to response bias.

Finally, Lindsay & Mitchee (1988) examined the reliability of different response formats with 29 adults with mild to moderate learning disabilities using the Zung Self-rating Anxiety Scale. They used a graded presentation format and a simple yes/no

answer format. Only the latter gave acceptable test-retest reliability ($\alpha=0.83$) and a significant split-half reliability correlation ($r=0.69$).

All the above studies deal with assessment of disorders either using previously developed scales (e.g. Lindsay *et al*, 1994) or through developing scales for this client group using existing criteria (e.g. Reynolds & Baker, 1988). All the studies questioned adults with mild to moderate learning disabilities, yet the results are somewhat discrepant. Reynolds & Baker (1988) found that their sample were able to use graded formats with acceptable test - retest reliability, a finding disputed by Lindsay & Mitchee (1988) who state that only their dichotomous presentation gave adequate reliability. Where studies have included measures of a subject's ability to use a scoring format reliably and validly (e.g. Helsel & Matson, 1988; Reynolds & Baker, 1988) no information is given about those participants who fail these screening tests. Therefore, it is unclear whether these participants are significantly different from those who pass the screening tests on measures which are hypothesised to promote reliable and valid responding (such as level of receptive vocabulary).

One recent study has broadened the area of research away from symptoms to opinions held by adults with learning disabilities. Dagnan & Ruddick (1995) examined the reliability of using visual analogue and personal questionnaires with adults with mild to moderate learning disabilities to assess their satisfaction with services offered by their local community teams. 29 adults were tested with a personal questionnaire, scored using a three-point response format, and a visual analogue scale consisting of a line anchored between two pictures illustrating the extreme points of the scale. The visual analogue scale had acceptable test - retest reliability for two of the three questions assessed, and the personal questionnaire was used consistently by the majority of participants. There was a significant effect of verbal ability (as assessed by the British Picture Vocabulary Scale: BPVS, Dunn, 1982) on the consistency of use of the interview format. Those with higher BPVS scores used this format more consistently.

This result was not replicated in the visual analogue reliability data, suggesting that this format may be more suitable for use with people with lower verbal abilities.

In summary, the ability of adults with learning disabilities to respond reliably and validly has been most researched in the assessment of depression. Results are somewhat inconsistent. However, all groups use a number of common strategies to maximise the likelihood of reliable responding. These tactics include simplifying response formats' wording and augmenting them with visual cues; presenting scales orally and individually and attempting to validate findings against other questionnaires or clinical interviews. Receptive vocabulary is considered a major limiting factor in an informant's ability to use scoring formats reliably. This may be attenuated if visual cues are used to aid understanding.

Reliability of self-report of children with learning disabilities

There are few reports of the reliability of self-report of children with learning disabilities. Those which exist tend to examine concepts other than diagnostic categories, such as "loneliness" (Williams & Asher, 1992) or "fears" (King, Josephs, Gullone, Madden & Ollendick, 1994).

Williams & Asher (1992) investigated whether the concept of loneliness is meaningful to children with learning disabilities by comparing the results of a loneliness questionnaire with a "Concept of Loneliness" interview. They tested 62 children with mild learning disabilities aged 8 - 13. The questionnaire was scored with a three point response format. The authors give no data on the reliability of using this scoring scale, but derive a loneliness questionnaire from the 10 items in the original questionnaire which had high factor loadings in both the sample with learning disabilities and the non-learning disabled comparison group. This scale was derived at the expense of dropping items which loaded heavily on the primary factor in the group with learning

disabilities only. This suggests that the concept of “loneliness” may be different for children with and without learning disabilities.

King *et al* (1994) investigated the fears of children with a range of disabilities (intellectual and sensory) using a previously developed instrument, the Fear Survey Schedule for Children - Revised (FSSC-R, Ollendick, 1983). They tested 302 children with mild to moderate learning disabilities, a sample which included children with syndromal causes of their learning disabilities and those of unknown aetiology. They were aged seven to 18. The children with learning disabilities were thought to “represent the greatest challenge in fear assessment”. The scale was administered individually to each child, with items read aloud. A visual analogue scale was used to enhance the three point fear rating scale used. King *et al* (1994) report 100% inter-rater reliability using this format and also assessed the children’s tendency to acquiesce by including two neutral items. These consistently received the lowest fear rating, leading to the conclusion that response acquiescence did not occur in their sample. Although the researchers did not report on test-retest reliability, they found that the FSSC-R had good internal consistency.

Sarphare & Aman (1996) examined fears reported by children with mild learning disabilities. They advocate using structured instruments in assessment, as there is a tendency for young children, and older children with learning disabilities, to either exaggerate their fears or be reticent if asked to report their fears in an unstructured way (Millar, 1983). The responses of children with learning disabilities may also be strongly affected by response sets and situational and capacity variables (Matthews & Levy, 1961). The children in this study were given the FSSC-R (Ollendick, 1983) and the Social Anxiety Scale for Children (SASC; La Greca, Dandes, Wick, Shaw & Stone, 1988). The FSSC-R was scored on a three point scale. No information is given about the scoring format used in the SASC. The test-retest reliability for children with learning disabilities ranged from moderately good to excellent, with correlations between Time 1 and 2 presentations of between .68 and .84. There were

highly significant correlations between all the subscales of the FSSC-R and all the subscales of the SASC.

Silon & Harter (1985) examined the validity of using a number of self-assessment instruments designed for use with normal IQ students with students with mild learning disabilities. They chose a structured alternative response format to reduce the tendency of this group to give socially desirable responses. This response pattern is particular problem if using yes/no response formats. Using the structured alternative response format gave internal consistencies for each scale of .75 to .86 and test-retest reliabilities of .69 to .80 after a test-retest delay of three months. A consistent finding from their study is that the concepts examined (e.g. "perceived competence") have a simpler factor structure in the learning disabled population than the normal IQ group. The items which load most highly on these factors are those which were worded very concretely and describe specific behaviours.

Few studies have examined those factors which affect reliable responding in children with learning disabilities. However, a number of points have been proposed which may be particularly important in their assessment, in addition to the strategies used in assessing adults with learning disabilities. These are that assessments should be structured to minimise the likelihood of exaggeration or reticence (Millar, 1983), and that situational and capacity variables may be major limitations on the responses given. There is evidence that the factor structure of concepts used with people without learning disabilities may not be appropriate when used with people with learning disabilities: in this group the factor structure is likely to be less differentiated (Silon & Harter, 1985). Finally, the content of scale items may need to be modified before instruments can be used.

Rationale for current study

This study was a preliminary investigation of the reliability of use of visual analogue and Likert response formats by children with mild learning disabilities. Studies to date with this group have used very simple response formats. It was the intention of this study to see if more differentiated scales could be reliably used, and to investigate some of the factors (such as level of receptive vocabulary) which might affect reliability of responding. The reliable use of more differentiated response formats is important if questionnaires are to be used to measure subtle changes in feelings or beliefs over time. The scale used had not been validated for use with this population, therefore its reliability had to be established in the first instance. Due to project constraints (only one researcher was available to administer the questionnaire) it was decided to investigate test-retest reliability.

The scale chosen to investigate the reliability of these response formats was the Body Esteem Scale (BES; Franzoi & Shields, 1984). Body esteem is an important component of self-esteem (McCaulay, Mintz & Glenn, 1988). In studies with a normal population the BES has been shown to correlate highly with measures of self-esteem (Franzoi & Shields, 1984). It was intended that the relationship between body esteem and self-esteem in this population would be investigated once the reliability of the BES had been established.

Hypotheses

The main hypothesis was that children with mild learning disabilities would be able to use five-point Likert and visual analogue response formats reliably. It was hypothesised that children with a higher level of receptive language ability would be more reliable than those with a low level of receptive language in using the Likert scale, but that this would not affect reliability of responding on the visual analogue scale (Dagnan & Ruddock, 1995). A further hypothesis was that children with learning disabilities would be prone to forming response sets (Matthews & Levy,

1963), therefore a condition was included to investigate this possibility. The final hypothesis was that children with learning disabilities would not use the two response formats comparably: that is a score of one on the Likert scale would not be equivalent to a score of one on the visual analogue scale. If true, one would expect to find larger discrepancies between the same question asked on Times 1 and 2 using a different format than in conditions where these questions were asked using the same response format.

Ethical approval

Ethical approval for the study was first obtained from the Assistant Director of Pupil Services at West Sussex County Council. The Head Teacher of a local school for children with moderate learning difficulties¹ was approached, and his permission obtained to ask Year 10 students to participate. Next, the form tutor of the Year 10 students was approached. Consent forms and information about the study were then sent to all parents of the Year 10 students. Finally, students were asked individually on each occasion if they would consent to take part. It was made clear that they were under no obligation to take part, and did not have to answer any questions about which they felt uncomfortable.

Method

Measures

The receptive vocabulary of each pupil was assessed using the BPVS (Dunn, Dunn, Whetton & Pintillite, 1982). This is a simple measure of receptive vocabulary which requires the subject to indicate which of four pictures best illustrates a given word. The BPVS is widely used in research with people with learning disabilities as it is

¹ Moderate learning difficulties used in an educational setting is equivalent in meaning to mild learning disabilities used in a clinical setting.

quick to administer and covers a wide range of abilities, therefore making it less susceptible to floor or ceiling effects (Dunn *et al*, 1982).

The BES was chosen to investigate the ability of children with learning disabilities to use Likert and visual analogue scales reliably. It was selected for a number of reasons. First, it was intended that, if reliable, the scale would be used in further research projects. Secondly, it is structured and objectively scored. Finally, it is the only body image questionnaire which has reasonable reliability and validity in normal populations (Wylie, 1979). In its original form it consists of 35 items scored on a five point Likert scale.

The BES was derived from the Body-Cathexis Scale (Secord & Jourard, 1953) which assumed that body image was a unidimensional construct. However, Franzoi & Shields (1984) discovered that, for their collegiate population, body image was a multidimensional construct. For their population the scale had a three factor structure, which was different for each sex. For males, body esteem was found to consist of factors of "physical attractiveness", "upper body strength" and "physical condition", whereas for females the factors were "sexual attractiveness", "weight concern" and "physical condition".

For the purposes of this study, the BES was assumed to measure body esteem as a unidimensional construct. This is a strong hypothesis and is justified by the following reasons. First, the group of students used by Franzoi & Shields were older than this test group, and it can be assumed that they were of above average intelligence. Therefore, there is no a priori reason to assume that the same factor structure would be valid for this population. Secondly, even in Franzoi & Shields' group there were strong correlations between the three factors of Body Esteem for each sex, suggesting that there might be one factor underlying the three. Finally Fisher (1986) suggested that there is a developmental component to body image, and that children and adult's body image may be significantly different. One feature of the development of body

image is that there is a gradual differentiation of different aspects of body image, hence children with learning disabilities may not have as differentiated a body image as college students of above average intelligence. There is evidence that the assumption of less differentiation in children with learning disabilities' self-concepts is warranted (Silon & Harter, 1985).

The scale was adapted for use as follows. The scale item wording was simplified by putting all the original items into simple statements phrased in the first person (e.g. "body scent" became "the way I smell"). Abstract notions were rephrased into concrete examples. These rephrasings were done in conjunction with the students' form tutor to try to ensure that the new item would be understood by the students and were accurate reflections of the original BES item. The original BES wordings and their modifications for this study are given in Figure 1. All questionnaires were administered individually with the items read aloud. Understanding was checked by asking participants to indicate, on their own bodies, the item being asked about, or by using materials drawn from the "Not a Child Anymore" information pack. Two scale presentations were used: the five point format was retained for half the questions but the wording was simplified from "having strong positive feelings" to "really like". A visual analogue format was also used: this consisted of a five inch line anchored between a "smiley" face and sad face. This is similar to the format used by Dagnan & Ruddick (1995), which was shown to be used reliably by adults with learning disabilities.

To address the hypotheses set out, the scale was administered as laid out in Table 1. The 35 questions were subdivided into four blocks, the first three consisted of nine questions each, the last of eight questions. There were four different presentation formats: Likert and visual analogue scales presented forwards and backwards. These four conditions are illustrated in Table 2. Thus four different versions of the BES were used, with each student being exposed to each condition of responding. This complexity of design was adopted to control for possible effects of order of

presentation. In addition, the students' form tutor had expressed concern that they might get bored if asked to answer questions all in one format or another, and suggested that the response format be varied to prevent this from happening.

The Likert format was scored from one to five with a score of one corresponding to the answer "really like" and five indicating "really don't like". The visual analogue scale was scored by dividing the five inch line into inch-long segments and scored one to five (again one indicating that they really liked the aspect being asked about, five indicating that they really disliked the aspect being talked about).

Figure 1

Original BES item wordings and modifications

<i>Original</i>	<i>Modified version</i>
body scent	the way I smell
appetite	the amount I eat
nose	my nose
physical stamina	how far I can run
reflexes	how fast I can move
lips	my lips
muscular strength	how strong I am
waist	my waist
energy level	how energetic I am
thighs	my thighs
ears	my ears
biceps	my arm muscles
chin	my chin
body build	the way my body is built
physical co-ordination	how well I can do two things at once
buttocks	my bottom
agility	how agile I am
width of shoulders	how wide my shoulders are
arms	my arms
chest or breasts	my chest
appearance of eyes	how my eyes look
cheeks/cheekbones	how my cheeks look
hips	my hips
legs	my legs
figure	my figure
sex drive	my sex drive
feet	my feet
sex organs	my private parts
appearance of stomach	the way my stomach looks
health	my health
sex activities	activities to do with sex
body hair	the hair on my body
physical condition	my physical condition
face	the way my face looks
weight	how heavy I am

Table 1

The order of BES presentation given on test and retest

Big Group	Small Group	time one qu1-9	time two qu1-9	time one qu10-18	time two qu10-18	time one qu19-27	time two qu19-27	time one qu28-35	time two qu28-35
A	a	Likert forward	Likert forward	Likert forward	Likert reversed	Analogue forward	Analogue forward	Analogue forward	Analogue Reversed
	b			Likert forward	Analogue forward			Analogue forward	Likert Forward
	c	Analogue forward	Analogue forward	Analogue forward	Analogue reversed	Likert forward	Likert forward	Likert forward	Likert reversed
	d			Analogue forward	Likert forward			Likert forward	Analogue forward
B									

Table Two

Four different response formats used to present the BES

Likert forward

really like *quite* like don't like don't like *really*
or dislike don't like

Likert reversed

really don't like don't like *quite* like *really* like
don't like or dislike

Visual analogue forward

Visual analogue reversed

Procedure

Each student was seen individually in a side room separate from their classroom. It was explained that the study was to see what they thought about various aspects of their bodies, and that anything they said was confidential and would not be repeated to their teachers or other students. Any questions which they had were answered, and their verbal consent to take part in the project obtained. On the first session the BPVS was administered, and on the basis of this and the sex of the student they were allocated to a group. The BES was then administered. The first session typically lasted 15-20 minutes. There was a three or six week delay between test sessions. On retest the BES was administered again according to the schedule given in Table 1.

Participants

Parents of all students from Year 10 of the MLD LEA school were asked if they would be agreeable to their son or daughter to take part in the study. Of the whole year group of 24, 18 students were available and consented to be tested on two occasions. One female student (allocated to group b) was tested at Time 1 but was unavailable to be retested. One male student (allocated to group a) who was available to be tested on both occasions was clearly responding regardless of the content of the questions. His data have been excluded from the following analyses. The sex, BPVS raw score and mean age of each group is given in Table 3.

Table 3

Sex, BPVS raw score and Mean Age of participants in each experimental group

Big Group	Small Group	Number (Male: Female)	Average Raw BPVS score (SD)	Mean Age/Months (SD)
A	a	4 (2:2)	16.8 (2.9)	180.8 (3.0)
	b	3 (2:1)	18.3 (2.8)	184.0 (2.7)
B	c	5 (2:3)	19.4 (2.4)	181.2 (4.7)
	d	5 (3:2)	18.7 (2.8)	181.8 (4.1)

A Mann-Whitney U-test showed that group A and B were not significantly different in age ($U = 31$, $p > .05$) or BPVS raw score ($U = 21.5$, $p > .05$). Further planned comparisons were between Groups a and c and b and d. Mann-Whitney U tests were used to compare these groups on age and raw BPVS score. Groups a and c did not differ on age ($U=8.5$, $p > .05$) or BPVS raw score ($U = 4.5$, $p > .05$). Groups b and d did not differ on age ($U=7$, $p > .05$) or BPVS raw score ($U=6$, $p > .05$).

Results

To address main hypothesis concerning test-retest reliability, the students were essentially put into two groups (given in the first column of Table 1 and labelled A and B). To address the further issues of the comparability between scales and whether or not the students formed response sets, these groups were further subdivided into smaller groups (given in column two of Table 1 and labelled a, b, c and d).

The reliability of using these response formats with this population was examined in two ways. First, the test-retest reliability of participants using the Likert forward and visual analogue forward response formats was calculated by summing the scores of each subject across questions 1-9 and questions 19-27, and correlating their Time 1

and Time 2 scores using Spearman rank correlation coefficients. Secondly, the absolute magnitude of discrepancy between presentation of a question at Time 1 and Time 2 was calculated. Discrepancy scores give a real and direct measure of the test-retest reliability of a scoring format from Time 1 to Time 2. The average discrepancy scores were compared using Mann-Whitney U-tests, as due to the small numbers involved in the study it could not be assumed that data were normally distributed. The relationship between discrepancy scores and BPVS raw scores was examined using Spearman rank correlation coefficients.

The correlations between Time 1 and Time 2 performance is given in Table 4.

Table 4

Correlation between Time 1 and Time 2 performance using both presentation formats, and the significance of this correlation

Group	Likert-Likert correlation (significance)	Analogue-Analogue correlation (significance)
A (n=8)	.123 (p>.05)	.918 (p<.05)
B (n=10)	.682 (p<.05)	.848 (p<.05)

There were strong positive correlations between Time 1 and 2 presentations using the Likert format (.858) and the visual analogue format (.950) in Group B. Both correlations in Group B were statistically significant (p<.05). There was a strong, positive correlation in Group A for analogue-analogue presentation, but there was no evidence of any correlation between Time 1 and 2 presentation of the Likert format in this group.

To assess the reliability of using Likert and visual analogue response formats with this group the mean discrepancy between Time 1 and Time 2 for group A and B on

questions 1-9 and 19-27 were calculated. The criteria for reliable responding was set as a mean discrepancy for each subject across these blocks of questions of one or less (Dagnan & Ruddick 1995). These means are given in Table 5.

Table 5

The mean discrepancy scores for Analogue-Analogue and Likert-Likert presentation

Group	Likert-Likert Mean discrepancy score	Analogue-Analogue Mean discrepancy score
A (n=7)	0.57	0.82
B (n=10)	0.74	0.42

To examine whether the mean discrepancy scores were different between Likert-Likert and visual analogue-visual analogue presentation, a Mann-Whitney test was carried out between the average discrepancy scores for each block of questions. There were no significant differences between the discrepancy scores of group A and B on either questions 1-9 ($U = 24$, $p > .05$) or questions 19-27 ($U = 28$, $p > 0.05$).

The results were then collapsed across question blocks to examine the effects of test-retest interval and sex on the reliability of responding in the Likert-Likert condition or the visual analogue-visual analogue condition. A series of Mann Whitney U-tests were carried out, and the results presented in Table 6. There are no significant differences between the discrepancy scores for either scale presentation for either sex, or after a three- or six-week test-retest interval.

Table 6

The effect of test-retest interval and sex of informant on reliability of responding

Condition	Test-retest interval U value (p value)	Sex of respondent U value (p value)
Likert-Likert	31 (>.05)	27 (>.05)
analogue-analogue	34 (>.05)	24.5 (>.05)

Finally the effect of level of receptive vocabulary (as measured by the BPVS) on the test-retest reliability was examined for each scale presentation using Spearman rank correlations. The correlations between BPVS raw score and Likert-Likert discrepancy score was .304 ($p > .05$) and between BPVS raw score and visual analogue-visual analogue scale -.328 ($p > .05$).

The second hypothesis was that children with learning disabilities would be susceptible to forming response sets. The mean discrepancy scores for group a and c on questions 10-18 and 28-35 were calculated. In these presentation, each group was asked a set of questions at Time 1 with the scoring format in one direction (see Table Two). On retest, the direction of scoring was reversed. These results are given in Table 7.

Table 7

Evidence for response bias in using Likert and visual analogue response formats

Group	Likert forward - reversed Mean Discrepancy score	Analogue forward - reversed Mean discrepancy score
a (n=4)	0.75	0.78
c (n=5)	0.75	0.91

A Mann-Whitney U-test between the average discrepancy scores for questions 10-18 showed that there was no significant difference between group a (responding using the Likert format forwards on Time 1 and reversed on Time 2) and group b (responding using the visual analogue format forwards at Time 1 and reversed at Time 2). For this block of questions $U=8.5$, $p>.05$. For the second block of questions there was also no significant difference between the two conditions ($U=10$, $p>.05$). Collapsing the data across blocks of questions and comparing all the questions answered using the Likert format with all using the visual analogue format shows that there is no difference between the average discrepancy scores in the two conditions ($U=48$, $p>.05$).

The third hypothesis was that children with learning disabilities would not use Likert and visual analogue formats comparably, that is a score of one on the Likert scale would not be comparable to a score of one on the visual analogue scale. If true, one would expect to find larger discrepancies between the same question asked on Times 1 and 2 using different scale presentations than in conditions where the same type of scale was presented on both occasions. The mean discrepancy scores for groups b and d on questions 10-18 and 28-35 are given in Table 8.

Table 8

The comparability of Likert and Visual Analogue scales

Group	Likert - Analogue Mean Discrepancy Score	Analogue - Likert Mean Discrepancy Score
b (n=3)	0.93	0.80
d (n=5)	0.58	0.58

Due to the small numbers in group b it was not possible to analyse the discrepancy scores within blocks of questions. The data were collapsed across blocks of questions and the average discrepancy scores analysed by condition (i.e. Likert - visual analogue discrepancy scores were compared with visual analogue - Likert). A Mann-Whitney

U-test showed that there was no significant difference between the discrepancy scores in these two conditions ($U=10$, $p>.05$).

Discussion

This study was a preliminary investigation into the ability of children with mild learning disabilities to use graded questionnaire response formats reliably. There is increasing recognition of the need to investigate reliable methods of self-report for assessing the thoughts and feelings of children and adults with learning disabilities. People with learning disabilities suffer a higher rate of affective disorder than the general population (Sturmey & Sevin, 1993), and thus there is a need for clinicians to be able to reliably and validly assess their thoughts and feelings.

Previous research with adults and children had suggested that people with learning disabilities may not be reliable respondents when using differentiated response formats. Some suggest that only yes/no presentations give acceptable reliability (Lindsay & Mitchie, 1988), although this type of format has been criticised as it may be more likely to produce socially desirable responses (Silon & Harter, 1985). In addition, yes/no response formats may be less clinically useful, as they do not allow for subtle changes to be measured. Therefore, in this study, two response presentations which allow for more differentiated responding were examined: visual analogue and five-point Likert scales.

The studies reviewed all acknowledge that, in using scales derived from the general population there is a need to modify their structure and content. Modifications commonly made to structure include: augmenting scales with visual cues; administering the scale orally and individually and demonstrating the scoring prior to administering the scale. The main alteration to content is to simplify item wordings.

This study was designed with the above findings in mind, with the aim of investigating whether children with learning disabilities could reliably use response formats which would have clinical utility in monitoring change over time. The study included conditions which examined whether level of receptive vocabulary affected reliability of responding with either format, that is whether Likert or visual analogue scales would prove more appropriate for use with children with lower levels of receptive vocabulary.

Interpretation of results

1. Test-retest performance on the same instrument

The first hypothesis was that children with mild learning disabilities would be able to use five point Likert and visual analogue scales reliably. This was examined by correlating scores obtained by Group A and B on question 1-9 and question 19-27. The evidence for this hypothesis is mixed. Group B showed highly significant positive correlations between their Time 1 and 2 responses on both types of response format, suggesting that they were responding reliably. Group A, however, showed a strong correlation between their Time 1 and 2 responses on the visual analogue scale, but no correlation between their Time 1 and 2 responses on the Likert scale.

Inspection of the data for Group A on questions 1-9 showed that two participants had large discrepancies between their answers at Times 1 and 2. One of these had the lowest BPVS score (13) of the whole sample - it had been hypothesised that reliable use of the Likert scale would be most dependent on receptive vocabulary ability. The other participant did not seem to be remarkably different from the rest of the group on BPVS score. The lack of significant correlation between Likert presentation on Time 1 and 2 is therefore hypothesised to be due to two outliers, one of whose performance may have been influenced by his low receptive vocabulary ability. Without these two participants the correlation increases to 0.68 which suggests that the rest of the group

was responding reliably to these questions. The very high correlations in Group B exclude explanations for the lack of correlation in Group A arising from level of receptive vocabulary, subset of questions asked or response format used as these factors were constant between the two groups.

The second way in which test-retest reliability was measured was by calculating the absolute discrepancy between each question's score when presented on each occasion. Reliable responding is represented by an average discrepancy across blocks of questions of one or less. Both Group A and B had average discrepancy scores of less than one for question 1-9 and question 19-27 i.e. for both response formats. This suggests that they were responding reliably. There were no significant differences between the groups on either block of questions.

2. Test-retest reliability across different conditions

This study also examined whether children with mild learning disabilities were prone to forming response sets when using structured questionnaire formats; whether they used visual analogue and five-point Likert scale formats comparably and whether there was a relationship between their level of receptive vocabulary and degree of discrepancy using either response format.

There was no evidence that the participants were forming response sets when using either response format. Average discrepancy scores calculated from both response formats were less than one, and there was no significant difference between any of the scores. The participants also used the two response formats consistently: their average discrepancy scores when answering questions using one format at Time 1 and the other at Time 2 were all less than one.

Finally, there was no evidence that level of receptive vocabulary was related to average discrepancy score for either Likert or analogue response formats. For the

range of BPVS scores sampled there were small, non-significant correlations between average discrepancy and BPVS raw score for both response formats.

Summary

These results suggest that, when presented with appropriately modified materials, children with mild learning disabilities can provide reliable self-report. The strategies used to modify the questionnaire were to use structured questionnaires which ask specific questions as this may prevent the problems of reticence or exaggeration. The scale was presented individually, and checks on comprehension were built in to the presentation of the scale. Comprehension was aided by use of visual materials, both to illustrate the content of the scale and augment the scale presentation. Finally, the wording of standard questionnaires was simplified to compensate for limitations of comprehension. These modifications may be necessary to promote reliable responding.

Given these favourable conditions, the participants in this study were reliable respondents. Across each block of questions given their average discrepancy between Time 1 and Time 2 presentation was less than one i.e. within the criteria set for reliable responding.

Clinical implications

The study examined the use of differentiated scales with children with learning disabilities, as these may be more useful and appropriate in clinical practice than using simpler response formats such as yes/no presentations. The conclusion is that both five point Likert or visual analogue scales can be reliably used by children with mild learning disabilities. Therefore either response format can be appropriately used to assess their feelings and opinions using self-report measures. Conventional questionnaires may be appropriate, if modified as suggested above. Important issues

in the use of self-report with children with learning disabilities centre on their comprehension of the content of scales used and response format selected. It is appropriate to enhance understanding using visual cues and pictures, and to amend scale wordings to present the concept examined as simply and as concretely as possible.

It may be practicable to obtain reliable self-report from children with learning disabilities in a clinical setting using the modifications and changes in scale presentation presented above. However, it is necessary to demonstrate sensitivity to change using these formats if they are to be of practical use. Furthermore, whilst it may be possible to administer questionnaires in the manner described above during the course of a piece of clinical work, it is not practical for assessing large numbers of children. This is necessary to investigate the nature of concepts such as “body esteem” in this population.

Critique of experimental design

Although the results reported here are encouraging, there are several aspects of the experimental design which make firm conclusions difficult to draw. First, the study was overambitious in trying to investigate so many variables, given that the study sample was small. There were too few participants and items per condition to examine reliability using more powerful parametric techniques. Therefore, the data were analysed using non-parametric measures and an agreement measure (discrepancy). Due to the small numbers, results could be disproportionately affected by outliers.

However, few researchers are able to administer tests to large numbers of people with learning disabilities. For example, Dagnan & Ruddock (1995) and Lindsay & Mitchee (1988) administered their scales to only 29 participants. Indeed, given the need to administer measures individually, it is unlikely that many researchers would have the resources to test very large numbers of participants. The question of reliable use of

self-report formats may be best addressed in this population by undertaking a series of small scale studies, or by undertaking a long-term study where several cohorts of participants could be assessed. Certainly if this study were repeated, more definite conclusions could be drawn if the study contained only one group, to whom the whole questionnaire was administered in one format on both occasions. Questions of scale comprehension and response bias may be better addressed by developing screening tests which could be administered prior to the main test battery. Such tests have been used in the adult population (Irvin *et al*, 1977), it may be appropriate to develop a similar measure for children.

Secondly, involving students from a local school led to unforeseen problems in the running of the study. It had been hoped to retest all participants after the same interval: unfortunately due to timetable clashes this was not possible. Therefore, an unexpected extra factor of test-retest interval was introduced. In addition it was not possible to test all the Year 10 students: this would have increased the number of participants to 24. The numbers were also reduced as two students who provided data were excluded from the final analyses: one was not able to be retested, the other was clearly responding irrespective of content.

In conclusion, the findings from this study are limited by the complexity of design. Given the number of students who could participate in the study, too many variables were included in the design to investigate other issues of interest (such as the assumption that the scale used measured one dimension in this population). Some of these variables were introduced due to the difficulties in conducting research in a non-clinical setting, where other activities have to take precedence over the running of research projects.

Future directions for research

There are several reasons why this population may have been able to use graded presentation formats reliably when other populations have not. First, the BES does not ask about abstract concepts. Reporting on abstract cognitions or transient physical feelings may be more difficult than reporting on body parts and easily described bodily properties (e.g. strength, smell). Concrete items are more reliably understood and reported on than abstract concepts (Silon & Harter, 1985).

Secondly, the BES requires the subject to judge how much an item is liked at that instant, rather than to make a judgement over a period of time. Temporal judgements may be particularly difficult for people with learning disabilities, whereas the notion of degrees of liking something may be more familiar. This is one variable which needs to be investigated to delineate the limits of reliable responding of children with mild learning disabilities.

Finally the sensitivity of using these response formats needs to be investigated. An initial aim of this project was to investigate whether children with learning disabilities could use graded response formats reliably. Many concepts used in clinical settings are not appropriately assessed in a yes/no fashion, yet this type of scale format is recommended as being the most appropriate to assessing people with learning disabilities (Lindsay & Mitchee, 1988). However to be of clinical utility measures need to be sensitive to change.

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Research on Placement

**Report of a reminiscence group run for older adults on an inpatient
continuing care ward**

All names and identifying features have been anomalised.

Context of research

This is a report of a reminiscence group co-run by the author as part of the Core Clinical Placement working with Older Adults. The Clinical Psychology Service to Older Adults had input to community and inpatient services, the latter being to a unit of four wards (the Hawthorn Unit). The unit comprised two wards which were long-stay, one for functionally impaired older adults, and one which was conceptualised as an assessment ward, from which older adults would be transferred into community settings such as nursing homes. In practice, this demarcation was not strictly observed, with a range of clients present on each ward. The main service offered to the unit by Clinical Psychology was assessment to aid in the differential diagnosis of dementia from depression. However, the Clinical Psychology Service was keen to expand its remit and increase its presence on the wards. Hence, when the manager of Violet Ward (a continuing care ward) expressed an interest in having a reminiscence group the opportunity was taken to set up a group, which was to be co-run by two clinical psychology trainees on the placement at that time.

This report is based on the format suggested by Powell & Adams (1993). A brief review of the literature is followed by the concept and aims of this study. The design and procedure are described, and the results given and discussed. The implications of this study for future reminiscence groups are stated and modifications to future projects suggested.

Review - the use of reminiscence in groupwork with older adults

A recent review of the use of reminiscence with older adults concluded that: reminiscence may be a pleasurable and helpful activity for older adults; it has dubious value as a therapeutic activity but may be a pleasant diversion which leads to improved communication, and that the most positive effect for clients may arise from improved staff knowledge of individual clients (Thornton & Brochie, 1987). Improved communication promotes the formation of meaningful relationships between group members, and hence group cohesion may occur (Hunter, 1989). This may in turn lead to increased life satisfaction, by helping to resolve past conflicts with the benefit of

current perspective, and re-establishing a sense of self where this may have been diminished (Poulton & Strässberg, 1990) .

Most of the studies reviewed by Thornton & Brochie concern cognitively intact older adults. The potential for cognitively impaired older adults to engage in reminiscence groups, and enjoy the potential benefits of appropriate activity, improved communication and positive impact on staff have been relatively neglected (McKiernan & Yardley, 1990). In working with cognitively impaired older adults, McKiernan & Yardley concluded that their clients with severe cognitive deficits were able to engage and contribute in reminiscence groups, and that this demonstrated the potential for such groups to be a meaningful and appropriate stimulating activity. The greatest benefit to cognitively impaired older adults may therefore be to provide meaningful cognitive activity, and this in itself is a valid aim for this client group (Head, Portnoy & Woods, 1990; McKiernan & Yardley, 1990).

Concept and aims of the present study

A reminiscence group was run on a continuing care ward for cognitively impaired older adults at the request of the ward manager. She expressed interest in having more day activities for her clients. The main aim of the group was, therefore, to provide an appropriate activity for this client group. The specific aims of the study were: to monitor involvement of individual members in the group and observe factors which impeded or promoted their involvement; to see if the group increased their life satisfaction and to actively involve ward staff in the running of the group. Ward staff have good knowledge of the day to day behaviour of participants, and could provide useful information on the quality of their interactions within the group, compared to on the ward. Head *et al* (1990) report that a valuable aspect of reminiscence groupwork is in promoting more complex and lengthy interactions in the group compared to in standard day-care settings.

Method

Participants

The Ward Manager, who knew the clients and had previous experience of running reminiscence groups, was asked to select four clients whom she believed would benefit from participating in a reminiscence group. The four patients she selected are outlined in Table 1, using information gathered from their case notes. All had a diagnosis of dementia, were vocal and suffered from no significant sensory impairments. There were no specific inclusion or exclusion criteria for the group.

Table 1: Outline of group participants

Participant	Age	Medication	Other diagnoses
Mrs. P.	83	2mg stelazine b.d.	"mentally subnormal"
		10mg temazepam o.d.	history of "paranoid illness"
Mrs. N.	74	nortryptiline (no details)	depression
Mrs. R.	85	None	None
Mr. B.	80	None	None

Measures

The study used two formal measures to address the aims of the group: one to investigate factors which promoted or impeded involvement in the group, and one to measure whether participation increased overall life satisfaction. The involvement of each participant in the group was measured using the Involvement Scale (IS) (Bender & Morris, 1990). The IS is a measure of participation in the group, and was completed by the group leaders for participants after each group session. It is scored on seven dimensions which measure different aspects of group involvement. A high score indicates the greatest involvement in the group. The maximum score is 23. Details of the IS and its scoring schedule are given in the Appendix.

The change in life satisfaction of group members was assessed using the Life Satisfaction Index (LSI: Luker, 1979). This is a self-report measure of life satisfaction and has been shown to be sensitive to change in this population (Bennett & Maas, 1988; Haight, 1988). The LSI is a twenty item questionnaire which was individually administered in the ward. It asks a person to rate their satisfaction with past and current aspects of their life and takes about 20 minutes to complete. Each item is scored two (indicating satisfaction), one (not sure) or zero (indication dissatisfaction). The maximum score is 40, with a higher score indicating greater life satisfaction. No normative data are available. A copy of the LSI is given in the Appendix.

Design

A single baseline design was planned for this study, with each group member being given the LSI one week before the start of the reminiscence group, and in the week following the last group. The group intervention was four, one hour, weekly reminiscence group sessions. As recommended by previous studies (Head *et al*, 1990) there was a high therapist to client ratio (1:2) and materials were chosen for the group which were personally relevant to individual members (McKiernan & Yardley, 1990). To promote ward staff's involvement and ownership of the group, a member of staff was asked to attend each session.

Procedure

A week prior to the first group session each participant was visited on the ward. The nature of the group was explained to them, and they were asked if they would like to participate. An attempt was made to complete the LSI with each participant. In the week following the last session the LSI was readministered to those group members who were capable of completing it.

The group was run on four occasions over a period of five weeks. There was a one week break after the first session. For the first and last sessions both co-therapists were present. The second session was taken by the co-therapist and the third by the

author. The same room was used on each occasion. Due to staff shortages a staff member was only available for the first two group sessions. The group began with four members. After the first session Mr. B moved to a nursing home. The remaining sessions were run with three group members, although Mrs. P became restless in sessions two and three, and was taken back to the day room. After each group session the IS was filled out by the co-therapists.

The materials chosen came from a local day centre and "Recall" - a Help the Aged publication (Johnston, 1981). The content of the first group was determined by the co-therapists, with subsequent sessions using material which had been raised in the previous group. Session One covered the local area, and made use of photographs to stimulate memories. Session Two looked at leisure activities and used pictures of local parks. Session Three looked at family life and chores, using old-fashioned household items such as moth balls and a flat iron. The final session used a tape of music hall songs and big band standards and focused on here and now enjoyment.

Results

Mrs. R and Mr. B understood the purpose of the group and the LSI. They were both willing to participate in the group. Mrs. P was not able to understand the LSI, and did not understand the nature of the group. She was encouraged to attend each group, but left as soon as she indicated that she wanted to leave. Mrs. N was happy to talk and be read the LSI but her answers cannot be considered reliable. For example, in answer to question 10 "I feel old and somewhat tired" she replied "It's tired and it's old and it's started to cough". She did not seem to understand the explanation of the group but was content to participate.

The group ran for four sessions as described above. Participants' behaviour and involvement was discussed and rated by the co-therapists after each session. The group seemed to be an enjoyable activity for two of the remaining participants, who both attended and engaged willingly. It was not unequivocally enjoyable for the third participant, who attended willingly at first but only stayed for two complete sessions.

The attempt to stimulate meaningful conversation between participants was partially successful. Mrs. R was the most talkative and lucid of the group, and was able to use all the materials presented to discuss her memories. When Mr. B was in the group, he and Mrs. R were able to discuss and compare their memories. Mrs. N was talkative but her contributions were consistently rated confused or inappropriate. Mrs. P was verbal but very difficult to understand. Mrs. N and Mrs. P's contributions often required considerable interpretation by the co-therapists, but sometimes seemed to be appropriate. However, their contributions often caused Mrs. R to become frustrated and she was only able to engage with the co-therapists.

Of the materials used the household objects (session 3) promoted most involvement, as even members who typically gave confused contributions could demonstrate their use. This session was extremely lively and enjoyed by the members who participated. The local photographs were very stimulating and useful for the least cognitively impaired in the group, they elicited few memories from the more cognitively impaired participants. The scores for each participant on each dimension of the IS is given in Table 2, and the total IS score by group in Table 3. It can be seen that individual group members have a large impact on the involvement of others, indicated by Mrs. R and Mrs. N being most involved when Mrs. P was not in the group (session 3). The effect of changing

Table 2: Individual IS scores for each group member

Group member	Mrs. R				Mrs. N				Mrs. P			
Session number	1	2	3	4	1	2	3	4	1	2	3	4
Willingness to join group	3	3	3	4	3	3	3	3	3	3	1	3
Confusion	3	3	3	3	1	0	1	2	2	2	*	1
Energy levels	2	2	2	2	0	0	2	1	1	1	*	0
Type of reminiscence	4	2	4	4	0	0	4	0	2	2	*	0
Interaction/relationships	3	3	5	3	2	1	4	4	2	2	*	0
Interest / participation	2	2	2	2	1	0	2	2	1	1	*	2
Enjoyment	0	2	3	3	1	0	1	1	2	1	*	0

* category not scored as participant did not join group.

group leaders on the IS scores of individuals was less striking.

Table 3: IS score by session number for each participant

Mr. B and Mrs. R can be considered reliable respondents on the LSI. Unfortunately, pre- and post-data is only available for Mrs. R. This data is presented below in Table 4.

Table 4: Pre- and post-group LSI scores

Participant	LSI score before group	LSI score after group
Mr. B	12	not available
Mrs. R	1	6

Discussion

The first aim was to monitor involvement of participants and note factors which impeded or promoted involvement. Two participants' IS scores increased across the sessions (Mrs. R and Mrs. N), indicating an increase in involvement and engagement with the group process. The final participant (Mrs. P) could not be engaged in the

group. Her IS scores showed the opposite pattern to the other participants', with a decrease in involvement across sessions. Consistent with the hypotheses of Bender (1990) and Head *et al* (1990), individual group members had a powerful effect on the involvement of others. In this case, Mrs. R and Mrs. N were the most engaged with the group when Mrs. P was not present. Furthermore, many of the difficulties in involving participants stemmed from the very different cognitive abilities of the group members. This made it difficult to pitch the group appropriately. Sometimes engagement of individual members was only possible if each was spoken to by a different therapist, as they were not able to converse with each other. Clearly, this impedes the formation of cohesion between group members.

Secondly, we aimed to see if the reminiscence group increased participants' life satisfaction. Only one participant (Mrs. R) was able to complete the LSI on both occasions, and she showed an increase in LSI score from one to six, pre- to post-administration. In general, the LSI was found to be too complex for these severely impaired clients, and therefore neither a reliable or valid measure.

Finally, we aimed to actively involve staff in the running of the group. We were successful in getting the ward staff to nominate participants for the group, and a staff member was able to attend for half the sessions. Given that the impetus for the group came from the Ward Manager, it was disappointing that a member of staff was not available for each session.

Implications

Reminiscence groups may be a meaningful activity for *some* cognitively impaired older adults. It is important to select group members carefully, such that there is the potential for cohesion to occur. In our group, the greatest barrier to cohesion was the differing cognitive abilities of participants: reminiscence groups may be most effective when the members are of broadly similar ability (Head *et al*, 1990). This is not to imply that the more cognitively impaired cannot engage in groupwork, as shown by

Mrs. N's consistent increase in involvement over sessions, and positive and appropriate reaction to some of the materials used.

The differing responses of Mrs. P and Mrs. N, despite a similar degree of cognitive impairment, illustrates that groupwork may not be the activity of choice for all cognitively impaired older adults. "Taster" sessions, in which groups could be brought together for one session to investigate levels of engagement and the ability to tolerate the format of the group may be a way of tailoring groups to the individual needs of members.

The second aim of the group was to investigate if reminiscence increased life satisfaction. The main conclusion to be drawn from this study is that the LSI is not an appropriate instrument to be used with very cognitively impaired older adults. The structure of the scale required a relatively high degree of verbal ability from participants, which rendered it unreliable and invalid for our participants.

Finally, there may be more appropriate ways of involving staff in the running of the group. The method used to recruit participants would have been more successful if specific inclusion or exclusion criteria had been given, for example selecting people of broadly similar cognitive ability, or with similar past interests. The potential benefits of having a member of staff in the group is that they can provide information on the quality of interactions in the group compared to in the ward environment, that is to provide evidence that the group is better at involving participants than the standard day-care. Thus ward staff's presence is very important in determining the impact of the group, and were this group repeated the importance of having a member of staff should be made paramount in the initial planning of the group.

Conclusion

A reminiscence group was run for four sessions on an inpatient continuing care ward. Despite changes in the participants and group leaders, it was found that reminiscence was an appropriate and enjoyable activity for the majority of group members. The

most important factor in promoting coherence was to have members of broadly similar cognitive ability. This influenced the interactions of group participants and their ability to use different reminiscence materials. People with very impaired cognitive abilities may still benefit from groupwork, but others may find the demands of being in a group stressful. Short "taster" sessions may be appropriate to determine those who would be able to participate in groups. Finally, ward staff have important contributions to make to the planning and running of reminiscence groups. It is suggested that this is explicitly recognised when such groups are planned, and their inclusion in sessions made a high priority.

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Appendix

The Involvement Scale

Recording sheet for individuals engaged in group activity

Name of group:

Session number:

Group work:

Date:

Names				
Willingness to join group				
confusion/inappropriate contributions				
energy levels				
type of reminiscence				
interaction/relationships				
interest/participation				
enjoyment				

1. Willingness to join group: 0 too ill (I) or absent (A); 1 refused to join group; 2 needed persuading; 3 needed reminding; 4 Came along without prompting
2. Confusion/ inappropriate contributions: 0 Did not contribute anything; 1 Almost all contributions confused/inappropriate; 2 Some contributions inappropriate; 3 All contributions appropriate
3. Energy level: 0 Doziness frequent; 0 persistent restlessness; 1 Intermittent doziness; 1 Intermittent restlessness; 2 Appeared calm and relaxed
4. Type of reminiscence: 0 No reminiscence; 1 recall neutral events; 2 Recall positive events; 3 Recalled negative events; 4 recalled positive and negative events.

6. Interaction/relationships: 0 Rude/inconsiderate; 0 Monopolised the session; 0 disruptive; Said nothing; 2 Spoke only to leaders when prompted; 3a made spontaneous comments to one-on-one on particular; 3b spontaneous comments to staff; 3c spontaneous comments to one other member; 4 Spontaneous comments to other members; 5 Helped others take part.
6. Interest/participation (need not be verbal): 0 Little response/uncooperative; 1 Active participation when prompted; 2 active participation without prompting
7. Enjoyment: 0 Showed no enjoyment; 1 occasionally showed enjoyment; 2 enjoyed majority of sessions; 3 Thoroughly enjoyed session

Seating plan:

1. Reasons for any absences
2. Topics/ material used: usefulness (+, 0, -).
3. Themes
4. Any factors affecting individual group members participation

NAME: ADDRESS:

.....
.....

DATE: DATE OF BIRTH:.....

PSYCHOLOGIST/ ASSESSOR:

LSI Score:

Introduction:

I am interested in how satisfied you are with your life up till now. I am going to read you some sentences/I would like you to read some sentences. If you agree with a sentence say/tick under Agree. If you do not agree say/tick under Disagree. If you are not sure either way say 'I don't know' or tick under ? Please be sure to answer every question on the list.

LIFE SATISFACTION INDEX

Life Satisfaction Index A

	Agree	Disagree	?
1 As I grow older, things seem better than I thought they would be	-	-	-
2 I have had more chances in life than most of the people I know	-	-	-
3 This is the dreariest time of my life	-	-	-
I am just as happy as when I was younger	-	-	-
My life could be happier than it is now	-	-	-
These are the best years of my life	-	-	-
Most of the things I do are boring and monotonous	-	-	-
I expect some interesting and pleasant things to happen to me in the future	-	-	-
The things I do today are as interesting to me as they ever were	-	-	-
I feel old and somewhat tired	-	-	-
I feel my age but it does not bother me	-	-	-
As I look back on my life, I am fairly well satisfied	-	-	-

	Agree	Disagree	?
13 I would not change my past life even if I could	-	-	-
14 Compared to other people my age, I've made a lot of foolish decisions in my life	-	-	-
15 Compared to other people my age I like to take an interest in my appearance	-	-	-
16 I have made plans for things I'll be doing a month or a year from now	-	-	-
17 When I think back over my life, I didn't get most of the important things I wanted	-	-	-
18 Compared to other people I get down in the dumps too often	-	-	-
19 I've got pretty much just about what I expected out of life	-	-	-
20 In spite of what people say the life of the average man is getting worse not better	-	-	-

Large Scale Research Project

The executive function and theory of mind performance

of boys with fragile X syndrome

Assessment A40

Abstract

Fragile X syndrome is a genetically inherited cause of learning disabilities. People with the syndrome typically have a characteristic behavioural profile of abnormal social behaviours which are behaviourally similar to those seen in people with autism. This study aimed to test whether cognitive processes which have been hypothesised to underlie the social abnormalities in people with autism were also impaired in boys with fragile X syndrome. Eight boys with fragile X syndrome and eight with learning disabilities of unknown aetiology, matched on receptive verbal ability, age and with no diagnosis of autism, were tested on a battery of theory of mind and executive function tasks. Significantly more boys with fragile X syndrome failed the simplest theory of mind task, although this could be attributed to overall level of ability rather than group membership. No differences were found between the groups on any other measures used. A proportion of both groups failed first and second order false belief tasks. The performance of both groups on the executive function measure was at the floor of the test. At low levels of overall ability the performance of boys with fragile X syndrome and boys with learning disabilities of unknown aetiology may be more similar than they are different. The implication of this result for clinical interventions is discussed.

Introduction

Fragile X syndrome has been identified as the most prevalent known inherited cause of learning disabilities (Sherman, 1996). Recent estimates of its prevalence vary from 1 per 2, 000 (Turner, Webb, Wake & Robinson 1996) to 1 per 5, 000 (Jacobs, Bullman, MacPherson, Youings, Rooney, Watson & Dennis, 1993) depending on the population studied. The syndrome is caused by a genetic abnormality, with associated characteristic physical, behavioural and cognitive features, which have begun to be identified in people affected by fragile X syndrome (Hagerman, 1996).

Behaviours and cognitive processes which are associated with a given genetic anomaly may be described as its behavioural phenotype. The study of behavioural phenotypes is expanding rapidly, fuelled in part by advances in molecular genetic techniques available to examine and characterise syndromes more precisely (Flint & Yule, 1994). The rationale for their study is set out below, and the difficulties in their definition and measurement explored. The proposed behavioural phenotype of fragile X syndrome consists of abnormalities of cognitive functioning, speech and language anomalies, attentional deficits and impairments in social behaviour (Turk, 1992). This study aims to expand the current description of the behavioural phenotype of fragile X syndrome by examining aspects of social cognition in the syndrome.

The study of behavioural phenotypes of conditions which cause developmental disabilities is closely linked with the study of neuropsychological development. The rationale is that children who are quantitatively different in their neuropsychological development may shed light on aspects of development in normal children. Both fields of study are in their infancy, and the relationship between the two can be considered reciprocal and dialectic (Pennington & Smith, 1988).

This study investigates the impairments in social behaviour found in boys with fragile X syndrome, using developmentally appropriate measures of executive function and theory of mind. The study draws upon theories of social impairment derived from work with children with autism which conceptualise social impairments as arising from

theory of mind and executive dysfunction. Thus, the literature pertaining to models of frontal lobe and executive function in children will be reviewed. This will include a consideration of the link between performance on executive function tasks and theory of mind tasks.

Finally, fragile X syndrome will be described, with reference to the areas of study listed above. Its behavioural phenotype will be described briefly, with particular emphasis on abnormalities of social cognition which form part of the phenotype. The similarity of these behaviours to behaviours seen in individuals with autism will be reviewed critically, and the rationale for this study laid out in detail.

The study of behavioural phenotypes

Definition

There is no consensus on the definition of a behavioural phenotype. A phenotype is an observed pattern of behaviours or traits, and is the result of an individual's genotype and environmental influences. A behavioural phenotype may be defined as a pattern of behaviour which is present in many or most individuals with a given condition (Turk & Hill, 1995). Other authors take a more restrictive view, and argue that behavioural phenotypes should consist of behaviours which occur in almost every case of a genetic or chromosomal disorder, and rarely or not at all in other conditions, and that there should be a direct and specific relationship between the behaviour and the genetic or chromosomal abnormality which was its cause (Flint & Yule, 1994).

It can be argued that this view is too restrictive. For example, if the first of Flint & Yule's criteria were applied to physical phenotypes, it would not allow for characteristics such as short stature or prominent ears to be included in a phenotype, as these features are commonly found in the wider population too (Turk & Hill, 1995). In the case of behavioural phenotypes, patterns of cognitive impairment, rather than syndrome-specific cognitive deficits may underlie behavioural phenotypes (Pennington & Ozonoff, 1996). That a behaviour or cognitive process is impaired in a number of groups should not logically exclude it from being part of a behavioural phenotype.

Flint & Yule (1994) also require behaviours to be present in almost every case of a disorder, that is to be consistent across affected individuals. However, consistency is a continuous rather than a dichotomous variable (Bennetto & Pennington, 1996). It is extremely unlikely that each individual affected by a developmental disorder would show the same behavioural phenotype, as each is different in the vast majority of their genes and in their specific environmental influences. Characteristic behaviours or cognitive processes may only be identifiable as the central tendency of carefully controlled groups. Bennetto & Pennington (1996) suggest that criteria should be developed for acceptable consistency for a given behaviour to be considered as part of a behavioural phenotype.

The last of Flint & Yule's stringent criteria is that there must be a specific relationship between the behaviour and the genetic or chromosomal abnormality. Flint (1996) argues that "phenotype" carries the connotation that a behaviour is primarily genetically determined, and therefore should be reserved for behaviours which are relatively free of environmental influences (for example, stereotypic movements). In contrast, many behaviours which are associated with syndromes, such as excessive food consumption in Prader Willi syndrome, or aspects of social cognition in fragile X syndrome are likely to have a more complex origin and to be a product of genetic and environmental influences. The rationale for labelling these behaviours as part of the behavioural phenotype, therefore, is to assume that at least a component of the behaviour has a close relationship with its genetic determinants. If this assumption is not made, the term "syndrome specific behaviour" may be more appropriate, as it accurately defines the relationship between the observed behaviour and syndrome, but makes no assumption about the nature of that association.

Applying such criteria to the definition of a behavioural phenotype avoids the difficulty of categorising behaviours by their cause, which can then imply that a behaviour is unchangeable. An alternative formulation is that behaviours which form part of a phenotype require special interventions, based on a more sophisticated understanding of the processes which underlie them. For example, special educational techniques

have been developed for children with fragile X syndrome (Hodapp & Dykens, 1992) based on an understanding of their cognitive strengths and weaknesses. However, describing behaviours as “syndrome specific” rather than part of a “behavioural phenotype” markedly reduces the number of behaviours which can be considered as part of the phenotype because the vast majority of physiological mechanisms which link behaviours to genetic abnormalities are unknown (Turk & Hill, 1995).

An alternative set of criteria to apply to behaviours are provided by Bennetto & Pennington (1996). They suggest that a behaviour should be considered part of the phenotype if it fulfils criteria of discriminant validity, specificity and primacy, in addition to consistency which was discussed above. Discriminant validity is necessary to show that a particular behaviour or cognitive process is impaired in affected individuals, but that other areas of functioning are relatively unimpaired. The behaviours investigated cannot be part of a wider pattern of impairment. Specificity is closely related to discriminant validity. The behaviours must be specific to the genetic abnormality under study and not a non-specific effect of growing up with learning disabilities. It is therefore important to use comparison groups of other children with learning disabilities. Finally, primacy is concerned with the relationship of an observed behaviour to the genetic cause, and is similar to Flint’s criteria of their being a direct relationship between observed behaviour and genetic abnormality. Many behaviours observed in genetic syndromes are complex, and establishing whether they are primarily genetically caused, secondary to a more fundamental deficit or correlated with other deficits is difficult. This issue can be addressed in a number of ways, including longitudinal studies, improvements in genetic and behavioural measures and the use of animal studies (Bennetto & Pennington, 1996; Flint, 1996).

This paper will adopt a more liberal set of criteria than those advocated by Flint & Yule (1994) when considering which behaviours can be considered as part of a behavioural phenotype. Behaviours will be described as part of the fragile X behavioural phenotype if they are not part of a more global impairment, that is children with fragile X syndrome show equivalent performance to the comparison group on other measures. To ensure that deficits are specific and not common to all children

with learning disabilities, a comparison group of children with learning disabilities of unknown aetiology is used. Finally results will be presented as group effects thus accepting that a given deficit may not be present in each child with fragile X syndrome.

Why study behavioural phenotypes?

The impetus to study behavioural phenotypes comes from a number of sources. The recognition that there are characteristic behaviours and cognitive profiles which are reliably associated with given syndromes has led to an increasing awareness that educational and clinical interventions may need to be tailored to take this into account (Hodapp & Dykens, 1992; Sobesky, 1996). However, as discussed above, every individual with a given syndrome will not display the same pattern of strengths and needs. Therefore not all will benefit from the same interventions or techniques. This issue may become clearer as research identifies more precisely how genotypic variations are manifested as phenotypic behaviours.

Secondly, the study of behavioural phenotypes can give important information on the likely course of an individual's learning disabilities and therefore facilitate familial adjustment and planning (Turk, 1996). Early identification of a genetic cause of learning disabilities (which may be prompted by a child showing characteristic behaviours or physical features of the syndrome) can also help to prevent secondary handicaps.

Finally, the careful study of behavioural phenotypes may lead to the identification of links between genetic substrates and aspects of behaviour, from which we might learn about the biological basis of normal behaviour (Flint & Yule, 1994). As discussed above, as yet there is limited understanding of specific brain-behaviour links. There are also a number of difficulties in the methodology of studying behavioural phenotypes, and these will be addressed below.

Methodological difficulties in studying behavioural phenotypes

There are a number of difficulties in the study of behavioural phenotypes which are useful to review. The major difficulty is that, even in relatively common conditions such as fragile X syndrome, the number of affected individuals is small. Furthermore, fragile X syndrome has a complex genetic basis, and it is likely that some aspects of the phenotype may be specific to certain subforms of the genotype. Therefore participants should be matched on the molecular basis of their genotype. Unfortunately, as described above, behaviours are unlikely to be present in every individual with fragile X syndrome and may only be measurable as a central tendency of a large number of participants. So, as researchers are given the genetic information they need to define their participants precisely and hence examine links between phenotype and genotype more specifically it will become increasingly difficult to recruit sufficient participants for a large group study.

Secondly, there is an issue of how behaviours or cognitive processes should be identified and measured. The two main approaches are either to use existing standardised measures or to develop a measure specific to the population under test. The advantages of the former are that they tend to have good standards of reliability and validity, and may have extensive normative data for comparisons. However, the use of standardised measures can lead to false positive associations being drawn between conditions. For example, the similarity of behaviours in males with fragile X syndrome and those with autism led to the hypothesis that fragile X syndrome was a single gene disorder predisposing to autism (Flint, 1996). Conditions which may be similar on standardised behavioural measures may be quite different if more sensitive, fine-grained measures are used, and is later discussed with reference to the relationship between autism and fragile X syndrome. The alternative requires new measures to be developed on a syndrome by syndrome basis. Again, the small numbers of participants available may make it difficult to develop suitably validated measures.

Summary

Behavioural phenotypes are patterns of behaviour (which can be broadly defined to include cognitive processes) which are reliably associated with given genetic abnormalities. There is some debate as to which standards should be applied before a behaviour can be considered part of a phenotype: more restrictive definitions could limit their study whereas liberal definitions could result in phenotypes becoming overinclusive. For the purposes of this paper, behaviours will be considered as part of the phenotype if they can be shown to have discriminant validity, specificity and a degree of constancy. The issue of primacy, which requires longitudinal studies, is beyond the remit of this study.

The study of behavioural phenotypes can provide important information to families, clinicians and educators in addition to furthering molecular genetic and neurodevelopmental research. Information about the specificity of behaviours to genotypes may soon be invaluable in informing clinical and educational strategies.

Finally, the study of behavioural phenotypes is complicated by the relatively small numbers of affected individuals. As molecular genetic techniques become more advanced there is the ability to categorise individuals very precisely by their genotypes. The challenge to researchers, then, is to develop measures which are sensitive enough to discriminate different behavioural phenotypes. The two approaches usually adopted are to use existing standardised measures, or to develop new measures for each behaviour. The former has the advantage of using reliable measures, but may miss subtle differences between behaviours. The main disadvantage with the latter is that small numbers of participants may make it difficult to validate measures.

Developmental neuropsychology

It is useful to review aspects of developmental neuropsychological research, as this study uses measures and theories of psychological development derived from work with typically developing children, in addition to those with developmental disabilities such as autism.

Definition

Developmental neuropsychology is concerned with the relationship between neurological processes and structures, and behavioural and cognitive development. There is as yet no mature science of neuropsychological development (Bennetto & Pennington, 1996). The difficulties in studying neuropsychological development will be outlined below, and different paradigms for its study explored.

Methodological difficulties

Research into neuropsychological development occurs in the context of rapid change in brain structure (Middleton & Schwartz, 1995). The infant brain changes and develops at a rapid rate by cells dividing in the germinal layers forming neurones which migrate to the cortex where multiple axonal and dendritic connections are formed (Goodman, 1989a). The processes of cell division and migration are complete by birth, with connections made before birth and during infancy. Post-natally, some connections are lost as functions become more localised (Goodman, 1989a).

Therefore, as the infant brain is developing and changing rapidly, it is of primary importance to use developmentally appropriate measures. Often measures are used which are merely downward extensions from adult tests (Welsh & Pennington, 1988). These may not be appropriate, as adult tests are usually designed to assess deficits from normal functioning. In contrast, developmental neuropsychology assesses skill acquisition (Chelune & Thompson, 1987). Adult tests may also not be able to detect precursors to normal adult functioning, leading to the erroneous conclusion that certain processes do not emerge in any form until later in the life span (Welsh & Pennington, 1988). Finally, adult tests may lack sensitivity to detect developmental changes in functioning.

Research methods

This study is concerned with aspects of social cognition in boys with fragile X syndrome: impairments in social functioning have been linked to executive dysfunction. There are a number of research paradigms which have been employed to

investigate neuropsychological development in children in addition to studies of children with developmental disabilities: these include animal models and studies of head injured children. Therefore, these research paradigms will be reviewed below, with reference to studies which examine the development of executive functions in children. Executive functions are defined as the skills necessary for the maintenance of an appropriate problem-solving set for attainment of a future goal, and include abilities such as set-shifting, set maintenance, interference control, inhibition, integration across space and time, planning and working memory (Pennington & Ozonoff, 1996). They are distinct from sensation, perception and aspects of memory and language, but have some overlap with attention, reasoning and problem-solving.

Animal models

Primates can be used to investigate the development of certain, limited behaviours during childhood. They have the advantage of being able to use close analogues of human developmental tasks in animal experiments. For example, human infants below 12 months of age have been shown to make an “A not B” error on a task requiring object-permanence. A toy is hidden in one of two locations, in full view of the infant, and after a brief delay they are allowed to search for it. Following a correct trial, the location of the toy is reversed. The “A not B” error refers to the child continuing to search for the toy in the previously correct location. The error is thought to reflect deficiencies in several processes including set maintenance over a delay (i.e. remembering the correct location of the toy) and inhibition of the prepotent response (i.e. reaching to the previously correct location) (Welsh & Pennington, 1988). These behaviours are both clearly included in the definition of executive functions.

Diamond & Goldman-Rakic (1986) investigated the performance of infant monkeys on the same object search task, and found the same developmental progression in ability on this task. They also found that adult and infant monkeys with frontal lesions made the same A not B error, whereas the performance of monkeys with parietal or hippocampal lesions was unaffected.

Thus animal models can be useful in investigating the development of executive functions where analogues of human developmental tasks exist. There is a difficulty in defining which cognitive processes are necessary for correct performance, and so converging evidence from a number of paradigms needs to be sought (Welsh & Pennington, 1988). Animal models have the advantage of being able to link brain areas and performance in the nonhuman primate. Non-invasive techniques for examining the areas of the human brain active during such tasks can be developed to investigate whether the same link holds for human performance. The main limitation on the use of animal models is that the tasks which can be used are quite restricted. Any task which requires verbal mediation obviously cannot be modelled. More complex tasks of social cognition, which are hypothesised to involve executive functions, cannot be investigated using this methodology.

Head injury

Head injuries, whether from external causes such as accidents, or internal such as stroke and tumours, have been extensively used in the study of adult neuropsychology. Children's head injuries provide less information about developmental neuropsychology for a number of reasons. First, focal lesions are more rare in childhood (Pennington & Ozonoff, 1996) and hence specific lesion-behaviour relationships difficult to evaluate. Those studies which have been published are single-case designs, and suffer from possible ascertainment biases as well as the lack of consistent and systematic measures used across cases. Secondly, any study of child neuropsychology has to take into account the developmental stage of the child at the time of injury (Oddy, 1993). This further limits the number of children with different focal lesions who can be studied. Finally, there is a continuing debate about the degree of localisation of function within the child's brain and its capacity for recovery following trauma (Oddy, 1993). Children's brains are less specialised and more plastic than adult brains (Goodman, 1989b) and therefore childhood brain injury may not give rise to the same areas of specific deficit as seen in adult populations with comparable lesions. For example, if the left hemisphere is damaged early in life, children are still able to acquire normal language as long as their right hemisphere is intact (Vargha-

Khadem, O'Gorman & Watters, 1985). However, it has been shown that brain-damaged children show specific deficits compared to IQ matched normal controls (Benton, 1974) and children with different sorts of epilepsy show different patterns of deficit (Fedio & Mirsky, 1969). In these cases, the site of lesion associated with a specific deficit are more or less correlated with the pattern found in adults (Oddy, 1993). Even so, the small numbers of children with specific lesions limits the widespread use of this methodology to investigate brain-behaviour links.

Developmental disabilities

Increasingly, research with children with developmental disabilities is used to overcome some of these difficulties in delineating specific brain-behaviour links. Where disorders have known causes, and hence something is known of the underlying neurochemical disruption this can be correlated against behaviours or performance on cognitive tests. For example, Welsh, Pennington & McCabe (1987) examined the performance of pre-school children with early-treated phenylketonuria on measures of executive function derived from adult tests and developmental tests. They found that the children with early-treated phenylketonuria were impaired on measures of executive function compared to children matched on IQ, and that executive function deficits were correlated with high levels of phenylalanine in the blood. This is considered to be a correlate of depressed dopaminergic functioning (Krause, Halminski, McDonald, Dembure, Salvo, Friedes & Elsas, 1985) and has been linked to executive function deficits in disorders with suspected frontal lobe pathology (Taylor, Saint-Cyr & Lang, 1986). Where the neuroscience of a disorder is well understood, paradigms such as this can be used to illuminate brain-behaviour links.

Summary

Animal studies, case reports from brain injured children and studies of children with developmental disabilities have all been used to investigate the neuropsychological development of executive function in childhood. Animal studies have used analogue tasks to show a similar pattern of performance between primates and human infants, and have shown that lesions to the frontal lobe depress performance on these

purported executive function tasks. However, animal models can not be used to investigate more complex executive function tasks, involving verbal mediation or social interaction.

Executive and frontal lobe functions in childhood

A definition of executive functions has been given in the previous section. Much complex human behaviour requires some aspect of executive function, and social discourse and social behaviour can be considered within the executive function domain (Dennis, 1991; Pennington & Ozonoff, 1996). Hence, it is suggested that the social impairments which are found in individuals with fragile X syndrome may be usefully investigated by examining aspects of executive function in this population.

Executive functions are descriptions of behaviour. One of the purposes of developmental neuropsychology is to investigate links between measures of behaviour or cognition and the brain mechanisms or structures which are presumed to underlie them. In adult neuropsychology, deficits in executive function are normally linked to deficits in behaviour seen in people with documented frontal lesions (Pennington & Ozonoff, 1996). There are a number of studies which have similarly linked performance on executive function tasks with frontal functioning in childhood, suggesting some utility of the “frontal metaphor” when examining executive functions in children.

First, as described above, Diamond & Goldman-Rakic (1986) found similar deficits on developmentally appropriate tasks of executive function when these tasks were administered to young children and primates. These deficits were specifically linked to frontal lesions in the primates, from which it can be inferred that this brain region is responsible for the behaviour in human infants (Welsh & Pennington, 1988). Thus, this study illustrates that aspects of executive function emerge early in life, and can be linked to frontal lobe function.

Secondly, there are some case studies which have documented the cognitive and behavioural effects of childhood frontal lesions. These studies may suffer from ascertainment bias, that is there may be a large number of children with similar frontal lesions who do not show behavioural effects. However, those which have been reported show that the effects of frontal lobe lesions in childhood are not silent, temporary or different from those seen in adults (Pennington & Ozonoff, 1996). This accords with the view that specific functional deficits related to lesions of a given location observed in children tend to follow the same pattern as those found in adults (Oddy, 1993).

Pennington & Ozonoff reviewed three syndromes affecting social behaviour which are commonly found following adult frontal lesions - akinetic mutism, pseudodepressed and pseudopsychopathic syndromes. Of these, the pseudopsychopathic syndrome is most commonly seen in children with frontal lesions. The child case studies they reviewed also reported many deficits of executive function on cognitive testing, with behavioural evidence of difficulties with attention and temporal integration. Thus, frontal lesions in childhood produce behavioural, social and cognitive effects similar to those seen in adults. The frontal metaphor may be useful in describing these profiles when found in children.

Finally, researchers have begun to investigate a developmental perspective of performance on tests which measure executive function. Chelune & Baer (1986) took an adult test of executive function found to be sensitive to frontal lobe damage (the Wisconsin Card Sorting Test (WCST) (Heaton, 1981) and gave it to children aged six to ten. They found a linear developmental progression in measures from the WCST argued to indicate concept formation (Number of Categories Achieved), suppression of prepotent responses (Percentage of Perseverative Errors) and an inability to ignore extraneous stimuli (Failures to Maintain Set). Children aged six scored more poorly on each of these measures than older children. There was a linear progression in performance on the WCST with age. By the age of ten, children's performance was indistinguishable from adult norms (Heaton, 1981).

Based on this study, Chelune & Baer provide normative data on the WCST performance of children aged six to 12. They tested 105 normally developing school aged children on the WCST and the Peabody Picture Vocabulary Test (PPVT: Dunn, 1965). The PPVT gives a measure of overall cognitive ability. They found modest correlations between the number of categories achieved and PPVT raw score ($r=.47$, $p<.001$) and number of perseverative errors and PPVT raw score ($r=-.47$, $p<.001$). They did not examine the relationship between total number of errors and PPVT raw score.

Other researchers have investigated the performance of children aged seven to fifteen on a range of tests purported to measure frontal lobe functioning (Levin, Culhane, Hartmann, Evankovich, Mattson, Harward, Ringholz, Ewing-Cobbs & Fletcher, 1991). Levin *et al* examined the performance of children aged seven to eight, nine to twelve and thirteen to fifteen on tests derived from adult tests (e.g. WCST), measures derived from developmental cognitive literature (e.g. the California Verbal Learning Test for Children (CVLT) (Delis, Kramer, Kaplan & Ober, 1986) and measures adapted from techniques developed by Luria (1966), such as the Go - No Go task.

They found a similar developmental pattern of functioning to Chelune & Baer (1986) on the WCST. The performance of children improved between six and ten years. The performance of children aged 10-12 years was indistinguishable from adult norms on the WCST. Performance on the Go-No Go task was similar. This task requires subjects to make a response in the presence of a particular stimulus and to refrain from responding in its absence. It therefore provides a measure of problem solving and inhibitory control. The major reduction in false-positive errors (a measure of the ability to inhibit responses) occurred by the age of twelve, with a small and non-significant decrease in errors between 9-12 year olds and 13-15 year olds. Therefore it seems that performance on tasks requiring inhibition of prepotent responses and concept formation is equivalent to adult performance by the age of twelve.

In contrast, Levin *et al* found that children aged 13-15 were better than those aged 9-12 on tasks requiring utilisation of semantic material (e.g. performance on CVLT).

Older children were also more efficient at planning strategies than the younger age groups. Therefore some aspects of executive function develop more quickly than others and reach an adult level of performance by the age of 12. In contrast, other executive functions are still developing at this age, and may not reach an adult level of performance until later.

Whilst these studies suggest that the developmental changes in performance of children on these tasks relate to the development of frontal lobe functioning, others do not validate the frontal metaphor in children. For example, Chelune & Thompson (1987) gave the WCST to a heterogeneous sample of children referred for neuropsychological evaluation. Their group included children who had suffered neurological trauma, epileptic seizures and learning difficulties. They found that the neurologically impaired children performed more poorly than their normative sample, at both younger and older age groups. However, the neurologically impaired children were also lower in overall cognitive ability, leading Chelune & Thompson to conclude that the WCST had clinical utility in measuring overall cognitive functioning. When the clinical group were divided into those with and without documented CNS dysfunction, older children performed better on each WCST measure than younger children, with the exception of the Number of Categories Achieved variable. There were no differences between groups on any of the WCST measures when examined on the basis of their neurological status (i.e. known or unknown CNS dysfunction). Thus the WCST may be sensitive to discriminating developmentally impaired from intact children, but lack specificity to different types of impairment. The correlations between measures of overall cognitive ability and WCST dependent variables (Chelune & Baer, 1986) may account for the finding that the WCST is sensitive to overall level of cognitive ability.

Chelune & Thompson's conclusion is tentative, as they give no details about the neurological conditions of their sample, for example where the foci of the epileptic seizures were, or what proportion of their group with documented CNS damage had frontal damage, focal or otherwise. Without such information, their results are difficult to interpret. However, the WCST has been shown in several studies to lack specificity to detect focal frontal damage in adult populations (Heaton, Chelune, Talley, Kay, &

Curtis, 1993). Patients with documented frontal lobe lesions have also been found to perform normally on the WCST (Damasio, Tranel & Damasio, 1991). Finally, the WCST may tap many other abilities in addition to executive function. It has already been shown that performance on the WCST is sensitive to overall level of cognitive ability (Chelune & Thompson, 1987). In addition, performance on the test is likely to be affected by knowledge and salience of the concepts of colour, form and number used in the test. Performance on the test may look abnormal if participants try out rules which are too complex, and hence complete very few categories (Nelson, 1976).

This is not to imply that the WCST has no utility in investigating deficits in executive function in children. The variable "Percentage of Perseverative Errors" has been found to be sensitive to impairment in executive function (Pennington & Ozonoff, 1996). Rather, caution should be used when using poor performance on the WCST to infer frontal lobe dysfunction, especially if experimental groups have not been matched on overall cognitive ability.

Finally, measures such as the WCST may be useful in identifying deficits in certain executive function skills, but these deficits are not specific to any one developmental disorder. The goal of neuropsychological studies of developmental disabilities has been to find the primary cognitive deficit in each disorder. Therefore, either deficits on the WCST cannot be the primary cognitive deficit in any developmental disability, as these deficits are too widespread, or measures from the WCST are too global to reflect subtle differences in the profile of executive function deficits across disorders (Pennington & Ozonoff, 1996). Indeed, the finding of a deficit on a WCST measure is the first step towards examining why performance is poor. The wide-spread finding of deficits on the WCST highlights the need for fine-grained measures of executive function to be developed to examine profile differences (Dennis, 1991).

Social behaviour, theory of mind and executive function

Damage to the frontal lobes in adults is associated with acquired abnormalities in social behaviour (Pennington & Ozonoff, 1996), and it has been hypothesised that social

discourse and competent social behaviour is regulated by the frontal lobes (Dennis, 1991). Acquired impairments cannot be directly equated to developmental abnormalities (Bailey, Phillips & Rutter, 1996). However, the notion that tasks assumed to be sensitive to frontal lobe dysfunction may be impaired in developmental disorders which are characterised by abnormalities in social behaviour (such as autistic spectrum disorders) has been investigated. The research relating to aspects of executive function in autism will be briefly reviewed below. This will include a discussion of the “theory of mind” hypothesis of autism, and the relationship between theory of mind and deficits in executive function discussed.

Autism and the “Theory of Mind” hypothesis

Autism is a life-long developmental disability, characterised by social and language abnormalities and stereotyped repetitive patterns of behaviour (Bailey *et al*, 1996). The social abnormalities are characterised by a lack of social reciprocity and an inability to form close personal relationships (Pennington & Ozonoff, 1996). One theory which has been advanced to explain the behavioural abnormalities is that people with autism lack a theory of mind (Leslie, 1987). Theory of mind refers to the ability to impute mental states to self and others, and is necessary to appreciate what other people know, believe or feel. It is also necessary to predict the effect that one’s behaviour will have on others.

Leslie (1987) proposed that the ability to understand others’ mental states was due to an isolated cognitive mechanism, operational from about the second year of life, which was necessary to manipulate second-order representations (metarepresentations). This ability allows normal infants to engage in pretend play, and later allows for the understanding of others’ beliefs and desires. Pretend play is impaired in children with autism (Baron-Cohen, 1987), and children with autism have been shown to be impaired compared to children with Down’s Syndrome and mental age matched, typically developing children on “false belief” tasks. These initial findings have been replicated using a range of different tasks (reviewed in Baron-Cohen, 1993).

Executive dysfunction accounts of autism

Whilst theory of mind accounts have been a useful research paradigm to investigate the social impairments characteristic of autism, this has been at the expense of explaining the other behavioural abnormalities which constitute the syndrome (Happé, 1994). An alternative perspective for examining the behavioural abnormalities comes from studies of executive dysfunction in children with autism. In a review of studies which examine executive function tasks in children with autism (Pennington & Ozonoff, 1996), the vast majority have found deficits in standardised tasks of executive function (e.g. WCST) when autistic children are compared to comparison groups. The dependent variables of "Perseverative Errors" and "Categories Obtained" derived from the WCST gave a significant difference between autistic and comparison groups in ten studies which employed these measures. The average effect size² (pooled across total errors and perseverative errors) was 1.06, which is considered a large effect of both clinical and statistical significance (Cohen, 1992).

The relationship between theory of mind and executive function deficits in autism

The question then arises as to the relationship between theory of mind deficits and impairment on tasks of executive function. Three main possibilities have been considered: either theory of mind or executive function deficits are primary, and the other a consequence of this primary deficit (Bishop, 1993); deficits on both tasks co-exist and are unrelated (Goodman, 1989b) or deficits on both tasks are correlates of some other cognitive process (Ozonoff, Pennington & Rogers, 1991a).

Detailed discussion of these possibilities are beyond the scope of this paper. However, some key issues in discussion of the relationship between executive function and theory of mind deficits will be presented below. First, the question of primacy may be best addressed by longitudinal studies. These will be complicated by the paucity of developmental measures of early precursors of executive function or theory of mind.

² The effect size was calculated by dividing the difference in means of the two groups by their average standard deviation.

Parents of autistic children tend to express concern at their child's development at around their second birthday (Bailey *et al*, 1996) although early indicators of autism may be reliably picked up earlier using specifically designed checklists (the Checklist for Autism in Toddlers (CHAT) (Baron-Cohen, Allen & Gillberg, 1992). Therefore measures need to be developed which can be used from a very young age.

Secondly, any attempt to explain all the behavioural abnormalities of autism by executive function deficits alone are confounded by these deficits existing, in isolation, in groups with no impairment of social interaction (Welsh, Pennington, Ozonoff, Rouse & McCabe, 1990). Welsh *et al* (1990) examined the executive function performance of children with early treated phenylketonuria and found pervasive deficits on standardised tasks of executive function without deficits in social behaviour. Therefore, deficits in executive functioning alone are not sufficient to cause abnormalities in social interaction. However, it remains a possibility that the children with early treated phenylketonuria showed a different pattern or severity of executive function deficits than those seen in children with autism or that they have more subtle impairments in social cognition (Bishop, 1993).

Thirdly, recent studies have shown that theory of mind and executive function deficits may not be unique or specific to children with autism (Zelazo, Burack, Benedetto & Frye, 1996). Zelazo *et al* (1996) found that their sample of people with Down's Syndrome (whose mean chronological age was 22.7 years) were impaired on a card sorting task and three theory of mind questions when compared to a comparison group matched on mental age (whose mean chronological age was 5.9 years). In addition their performances on these two measures were correlated, suggesting that, in this sample, performance may be mediated by more general cognitive ability. This study also demonstrates that impaired performance on theory of mind tasks may not be sufficient to produce the abnormalities of behaviour seen in autism, because individuals with Down's Syndrome have a cognitive and behavioural profile that is quite different to people with autism (Zelazo *et al*, 1996).

This study is a challenge to the ability of executive function and theory of mind deficits to explain the behaviour seen in people with autism. However it suffers from a number of methodological problems. First, the card sort used was idiosyncratic, and was quite different in its demands to more commonly used measures such as the WCST. For example, the participants were given the rule to sort to, reducing the load on working memory hypothesised to form part of the task demands of the WCST (Pennington & Ozonoff, 1996). Secondly, the study used a mental age matched comparison group, which did not control for non-specific effects of growing up with a developmental disability (Bennetto & Pennington, 1996). The two groups were matched on the basis of verbal mental age only, and no attempt was made to assess their broader cognitive abilities. This is unfortunate, as some of the data presented suggests that a more parsimonious explanation of the differences found between groups was that the task demands were beyond the capabilities of the participants with Down's Syndrome. For example, on the card sort, four out of twelve participants with Down's Syndrome failed to learn the task at all, compared to none of the comparison group. Those in the comparison group were "nearly always correct" on the card sort. Thirdly, despite referencing a study by Baron-Cohen *et al* (1995) which claimed that theory of mind deficits were specific to children with autism when compared to children with Down's Syndrome, Zelazo *et al* (1996) make no mention of the differences in their findings. Finally, the participants with Down's Syndrome were not screened for autism. Although the co-occurrence of autism and Down's Syndrome is low (Howlin, Wing & Gould, 1995), it cannot be ruled out that some of the individuals in the experimental group had autism or an autistic spectrum disorder.

A final possibility for the relationship between theory of mind and executive function performance is that they are separate, co-existing deficits (Goodman, 1989b). The classic way to investigate this issue is to look for a double dissociation in performance on the two abilities, that is to look for two groups, one of which has intact theory of mind performance but impairment on executive function tasks with the other showing the opposite pattern of performance. Ozonoff *et al* (1991b) found a single dissociation in that their sample of high-functioning children with autism or Asperger's syndrome both showed impairments on executive function tasks when compared to typically

developing children, but the children with Asperger's syndrome showed no impairment on theory of mind tasks compared to the comparison group.

Summary

Deficits in social behaviour in adults have been linked to damage to the frontal lobe area. Frontal lobe pathology is often inferred in patients who show deficits in tasks measuring executive function. It is hypothesised, therefore, that appropriate social behaviour in adult populations is mediated by executive functions (Pennington & Ozonoff, 1996).

Findings such as these have been used to inform studies of the development of social behaviour in children. The most widely studied developmental disability with characteristic abnormalities in social behaviour is autism. Children with autism have been found to perform poorly on tasks requiring "theory of mind". It was hypothesised that these deficits alone could explain the behaviours characteristic of autism. However, researchers have found prominent deficits in executive function tasks in samples of people with autism, suggesting that deficits in theory of mind alone cannot be responsible for their pervasive behavioural abnormalities.

Thus, both "theory of mind" tasks (coming from research on children with autism) and executive function deficits (identified in adults with acquired abnormalities in social behaviour) have been suggested as abilities which are necessary for competent social behaviour in humans.

Summary of developmental neuropsychology

Developmental neuropsychology is the study of behaviours and cognitive processes, and their links to brain function. The study of developmental disabilities is becoming an important methodology in examining these links. Where something is known of the neuroscience of a disorder, profiles of neuropsychological performance can be tentatively linked to brain mechanisms (Welsh *et al*, 1987; Welsh *et al*, 1990).

Converging evidence from animal studies, case studies of head injured children and studies of children with developmental disabilities suggest that executive function impairments can be linked to frontal lobe pathology. However, the measures commonly used to identify executive function deficits are sensitive to other variables too (such as overall cognitive ability), and hence deficits in task performance cannot be automatically equated with frontal lobe dysfunction.

Finally, the relationship between social behaviour and executive function was examined, with reference to studies of the behavioural abnormalities in autism. Recent studies have shown that theory of mind performance may be correlated with performance on tasks of executive function in groups of children with autism (Ozonoff *et al*, 1991) and children with Down's syndrome (Zelazo *et al*, 1996). Deficits in executive function alone are not sufficient to cause the abnormalities in social behaviour seen in autism (Welsh *et al*, 1990) and indeed executive function and theory of mind deficits can be found in groups with no abnormalities in social behaviour (Zelazo *et al*, 1996).

However, theory of mind and executive function performance are a useful starting point from which to examine social behaviour. Theory of mind studies have been useful in examining the social deficits characteristic of autism, although less useful in examining other, non-social abnormalities in behaviour (Happé, 1994). Executive function deficits have been identified in adult populations with acquired brain damage leading to abnormalities in social behaviour and prominent executive function deficits have also been found in children with autism (Pennington & Ozonoff, 1996).

Fragile X syndrome

Fragile X syndrome is a developmental disorder which accounts for a high proportion of X-linked learning disabilities (Hagerman, 1996). Researchers have begun to identify a putative behavioural phenotype (Turk, Hagerman, Barnicoat & McEvoy, 1994) which includes characteristic abnormalities in social behaviour. These are not usually consistent with a diagnosis of autism. A recent estimate is that around 2.5% of people

with autism have fragile X syndrome (Bailey *et al*, 1996). A recent study estimated the prevalence of autism (using DSM-III criteria) (APA, 1980) in males with fragile X syndrome to be 7% (Bregman, Dykens & Watson, 1988). The genetic basis of the syndrome, and aspects of the behavioural phenotype will be described below, and the rationale for this study laid out.

Genetic basis of fragile X syndrome

Fragile X syndrome has a known, complex genetic basis. Individuals with fragile X syndrome inherit an X chromosome with a fragile site, usually from their mothers, but very occasionally from their fathers. Chromosomes are composed of DNA which itself consists of two chemical strands comprising pairs of "base elements" - adenine, guanine, thymidine and cytosine (A, G, T and C respectively). These bases code in threes ("triplets") for amino acids, which are the building blocks of proteins. Fragile X syndrome arises when a CGG triplet expands dramatically at the fragile site on the X chromosome (Fu, Kuhl, Pizzuti, Sutcliffe, Richards, Verkerk, Holden, Fenwick, Warren, Oostra, Nelson & Caskey, 1991). In the general population there is a variable number of CGG repeats at this fragile site. Between six and 54 repeats are commonly found, with 29 repeats being the most common number (Fu *et al*, 1991). In families affected by fragile X syndrome, family members can be differentially affected by the fragile X gene. Those with between 52 and 200 CGG repeats are said to carry the pre-mutation (Oberle, Rousseau, Heitz, Devys, Hanauer, Boue, Bertheas & Mandel, 1991). Those with over 200 CGG repeats are said to have the full mutation.

In addition to the number of CGG repeats there are other genotypic variables which may be important in the expression of the phenotype. These differ for male and female carriers of the fragile X gene. Males with over 200 CGG repeats are said to carry the full mutation. However, in addition to the abnormal expansion of the CGG sequence, there may also be changes in a neighbouring area called the CpG "island" which is thought to play a role in gene regulation. This island is usually unmethylated, but is usually abnormally methylated in the full fragile X mutation (Oberle *et al*, 1991). The effect of this methylation is to prevent the production of the gene product usually

associated with the fragile X gene. This difference in genotype may lead to phenotypic differences, which will be discussed below. Finally, males may be mosaics - that is, have the full fragile X mutation in some cells, but the pre-mutation in others (Hagerman, 1992). This pattern is associated with less profound cognitive impairments than those seen in males with the full mutation

The behavioural phenotype of fragile X syndrome

The behavioural phenotype of males with the full fragile X mutation consists of learning disabilities, abnormalities of speech and language, attentional problems and impairments in social behaviour. The focus of this study are the social impairments found in affected individuals, which are investigated using developmentally appropriate measures of executive function and theory of mind. Two features of the behavioural phenotype relevant to the study will be discussed in detail: the apparent decline in overall IQ with age and impairments in social behaviour. This will include a consideration of the difficulties in selecting appropriate measures when studying this group, arising from the instability of IQ in boys with fragile X syndrome.

Developmental decline in IQ

Early cross-sectional studies of the IQ trajectory of males with fragile X syndrome seemed to show that there was a dramatic decline in IQ from child- to adulthood. Typically boys with fragile X syndrome were found to have borderline to mild learning disabilities whilst adult males had moderate to severe learning disabilities (Lachiewicz, Guillion, Spiridigliozzi & Aylesworth, 1987). This is in contrast to the relative stability of IQ in children with learning disabilities of unknown aetiology (Silverstein, Leguti, Friedman & Takayama, 1992), and the variable trajectories seen in other aetiologically specific groups of learning disabled children (Dykens, Hodapp, Walsh & Nash, 1992; Hodapp & Zigler, 1990).

Recent studies have suggested that the apparent decline in IQ is not as dramatic as first thought. Methodologically sound studies, using longitudinal designs and the same assessment measures at times one and two have found that IQ scores do decline in

males with fragile X syndrome, with differences in IQ ranging from five to 14 IQ points (reviewed in Bennetto & Pennington, 1996). However, not all boys with fragile X syndrome show declines in their IQ scores. Instead this seems to be limited to a subset of boys. One study which measured the molecular status of the genotype (Wright-Talamante, Cheema, Riddle, Luckey, Taylor & Hagerman, 1996) found that the decline in IQ was restricted to those boys with full methylation of the CpG island neighbouring the fragile site.

Other researchers have attributed the decline in IQ to syndrome-specific differences in information processing styles. Dykens, Hodapp & Leckman (1987) noted that fragile X boys performed poorly on sequential relative to simultaneous information processing tasks. There is some degree of specificity to the profile as boys with Down's Syndrome did not show a difference between their performance on sequential and simultaneous processing tasks, and although some boys with learning disabilities of unknown aetiology showed a weakness in sequential processing this group had not been screened for fragile X (Hodapp, Leckman, Dykens, Sparrow, Zelinsky & Ort, 1992). Dykens, Hodapp & Leckman (1994) suggested that this may be a result of deficits in frontal lobe functioning, as this area has been nominated as primarily responsible for sequential information processing (Das, Kirby & Jarman, 1975).

Abnormalities in social behaviour

Males with fragile X syndrome show a behavioural profile which includes many features which are behaviourally similar to those seen in autistic samples. These include:

i) gaze aversion. It has been widely reported that males with fragile X syndrome show a marked aversion to eye contact, and will avert their gaze rather than look directly at someone else (Wolff, Gardner, Paccia & Lappen, 1989). This is very similar to the DSM-IV (APA, 1994) criteria for autistic disorder. However, fine-grained analysis has indicated that there may be important differences in the abnormality of gaze shown by these groups. Cohen, Vietze, Sudhalter, Jenkins & Brown (1989) report that boys

with fragile X syndrome with and without autism showed gaze aversion which was unrelated to their verbal ability or age. They were also sensitive to parent initiated gaze. In contrast boys with autism but without fragile X syndrome showed less gaze aversion with age and verbal ability and were not more sensitive to parental gaze.

ii) *social relatedness*. Researchers consistently find a pattern of socially avoidant behaviour in males with fragile X syndrome. For example, Kerby & Dawson (1994) compared nine men with fragile X syndrome and nine men with learning disabilities of unknown aetiology on the number of autistic features shown, adaptive behaviour, temperament and schizotypal and schizoid personality disorders. They found that the men with fragile X syndrome met more of the DSM-III-R (APA, 1987) criteria for autism, showed no difference in adaptive behaviour and showed more features associated with schizotypal and schizoid personality disorders. In investigating social behaviour the researchers separated social skills from socialisation i.e. knowing how to socialise from putting those skills in to practice. The men with fragile X syndrome did not differ in their knowledge of social skills but were significantly less sociable than men in the comparison group.

Thus males with fragile X syndrome show impairments in social behaviour when compared to males with learning disabilities of unknown aetiology which are similar on a behavioural level to the social behaviour of people with autism. Even when boys with fragile X syndrome and a diagnosis of autism are excluded from groups of boys with fragile X syndrome, there remains a behavioural profile of communicatory and stereotypic “autistic-like” disturbances (Turk & Graham, 1997). On a cognitive level, few investigations have been undertaken into cognitive processes underlying these social deficits. However, those which have support the notion of the social impairment in fragile X syndrome being quantitatively different from that in autistic disorder. Simon & Finucane (1996) found that males with fragile X syndrome were not impaired on facial emotional recognition when compared to males with learning disabilities of unknown aetiology, whereas emotional perception is consistently impaired in children with autism (Ozonoff *et al*, 1991a).

The similarities between the abnormal social behaviour in fragile X syndrome and that seen in people with autism initially led to hypotheses that fragile X syndrome could be an important genetic abnormality which predisposed to autism. The similarities in behaviours between the two conditions may have been overestimated by the use of standardised measures (Flint, 1996), whereas when more specific measures are used significant differences emerge between people with fragile X syndrome and those with autism. However, the finding that social abnormalities in males with fragile X syndrome are different to those in people with autism does not advance understanding of social behaviour in fragile X syndrome.

Rationale for current study

This study aims to investigate the reported deficits in social behaviour found in boys with fragile X syndrome. Although the abnormalities in social behaviour in fragile X syndrome can be distinguished from those in autism on a behavioural level, there remains a consistent finding of “autistic-like” behaviours in boys with fragile X syndrome (Hagerman, 1992; Turk, 1992; Turk & Graham, 1997). Therefore, the study will examine whether boys with fragile X syndrome show deficits on measures which have been used to investigate social impairments in people with autism, namely theory of mind tasks. In addition, their performance on measures of executive function will be studied, for three reasons. First, executive functions have been hypothesised to be important for competent social behaviour (Pennington & Ozonoff, 1996). Secondly, females with fragile X syndrome have been shown to be deficient on executive function tasks (Mazzocco, Pennington & Hagerman, 1993). Finally, boys with fragile X show a weakness in sequential information processing (Dykens *et al*, 1987), which has been attributed to frontal lobe dysfunction (Dykens *et al*, 1994).

As boys with fragile X syndrome have been shown to be quantitatively different from boys with autism on behaviour measures and some cognitive measures, it is hypothesised that they would show no deficits on theory of mind tests compared to an appropriately matched comparison group of boys with learning disabilities. Kerby & Dawson (1994) hypothesised that males with fragile X syndrome are impaired in their

ability to use social rules, rather than their understanding of those rules per se. If theory of mind tasks tap some aspect of understanding social behaviour it is hypothesised that boys with fragile X syndrome should not be impaired on theory of mind tasks when compared to boys with learning disabilities of unknown aetiology.

The second hypothesis is that boys with fragile X syndrome are impaired on tests of executive function compared to the comparison group. Executive function deficits are widely implicated in abnormal human social behaviour (Pennington & Ozonoff, 1996), and deficits in executive function tasks have been found in women with fragile X syndrome (Mazzocco *et al*, 1993).

This study aimed to investigate two questions: first do boys with fragile X syndrome differ in their performance on theory of mind and executive function tasks when compared to appropriately matched controls? Secondly are those differences primarily due to their group membership? An alternative hypothesis is that deficits in task scores are produced by a few participants scoring poorly on a given task; that is that deficient performance is not widespread amongst the group, or that poor performance is a function of another variable such as cognitive ability. Each task is therefore analysed first for group differences between measures, and secondly for the specificity of any impairment to the fragile X group.

Method

This project used two independent groups: boys with fragile X syndrome and no diagnosis of autism, and boys with learning disabilities of unknown aetiology and no diagnosis of autism.

Participants

Eight boys with fragile X syndrome and eight with learning disabilities of unknown aetiology were recruited from the following sources.

The boys with fragile X syndrome were drawn from a sample of children who had participated in an earlier research project (Turk, 1995). They had originally been identified from registers held in Thames Regional Health Authority genetic centres, and lived in and around the Greater London area. They had all been diagnosed fragile X positive cytogenetically using standardised procedures and had no significant physical illness or physical handicap. There was no other specific cause for their learning disabilities. As part of their assessment for Turk's research they had completed the short-form British Ability Scales (BAS) (Elliot, Murray & Pearson, 1982) and been screened for DSM-III-R Autistic Disorder using the Schedule of Handicaps, Behaviours and Skills (Wing, 1980). For inclusion in this study the following criteria were applied:

1. aged 10 - 16 years
2. no diagnosis of DSM-III-R Autistic Disorder
3. a predicted IQ, based on their previous assessment using the BAS, of 50 - 70.

From Turk's original sample 13 boys with fragile X syndrome were identified as fulfilling the inclusion criteria for this study. These were approached and asked if they would be agreeable to participate in this study. From the 13 identified, all eight who replied consented to participate in the study.

It was intended to recruit further participants for the fragile X group from the local Child and Adolescent Developmental Disabilities Team, which regularly receives referrals of boys diagnosed with fragile X syndrome. During the period of the study no boys who fulfilled the inclusion criteria were referred. Further participants were then sought from paediatricians in the Greater London area who had shown interest in running joint clinics for their patients with fragile X syndrome. Unfortunately, due to resource constraints, it was not possible to include further boys with fragile X syndrome from this source.

A comparison group was approached from Turk's pool of participants. This group fulfilled all the criteria set out above, and in addition had had negative cytogenetic tests for fragile X. Thus the comparison group consisted of boys with learning disabilities of

unknown aetiology and no autism. 11 boys fulfilled the criteria for inclusion. These were approached and asked if they would be agreeable to participate in this study. Two consented to participate: they were excluded from the final sample as one was extremely high-functioning and the other extremely low functioning.

Further participants for the comparison group were then recruited from a local school for children with Moderate Learning Difficulties with established links to the Child and Adolescent Developmental Disabilities Team. The criteria for their inclusion in the study were:

1. aged 10 - 16
2. predicted IQ in the mild - moderate learning disabilities range
3. no known cause of their learning disabilities
4. no identified autism or autistic spectrum disorder, as noted on their Statement of Special Educational Needs (The Education Act, 1981).

Ten potential participants were identified by the school's Deputy Head. To avoid any potential recruitment bias as far as possible they were matched on date of birth with the participants with fragile X syndrome. These students' parents or guardians were approached and asked if their son would participate in the study. All consented to do so. The students were assessed at school, and eight who matched the fragile X group were included in the final sample.

Therefore, the final sample consisted of eight boys with fragile X syndrome recruited from Turk's study and eight boys with learning disabilities of unknown aetiology recruited from a local school for children with moderate learning difficulties.

Measures

Measures were chosen according to the following criteria. As far as possible, standardised assessment instruments were used with documented reliability and validity. Where standardised instruments did not exist (e.g. Theory of Mind tasks), tasks were utilised which had been commonly used in research, had well-documented

protocols for their administration and were suitable for children with cognitive abilities well below those predicted for the participants in this study.

Measures of overall cognitive ability

All participants were assessed using the Wechsler Intelligence Scales for Children - Third Edition (WISC-III^{uk}; Wechsler, 1992). The WISC-III^{uk} consists of ten subtests which measure different aspects of overall cognitive ability, and which combine to give two subscales of Performance IQ and Verbal IQ. The WISC-III^{uk} is widely used in clinical practice and research. Three dependent variables were taken from this measure: Performance IQ, Verbal IQ and Full Scale IQ. It was intended to match the groups on all three of these variables if possible. As there is some evidence that boys with fragile X syndrome have an uneven cognitive profile (Hodapp *et al*, 1992), with relative strengths in verbal abilities (Freund & Reiss, 1991) it was important to match groups on a wide range of variables.

After the first three fragile X participants had been tested it was realised that their IQ scores were significantly lower than had been predicted from Turk's research. The participants were scoring at or close to floor on the WISC-III^{uk}. A second measure was added to the assessment battery: the British Picture Vocabulary Scale (Dunn, Dunn, Whetton & Pintillie, 1982). This is a measure of receptive vocabulary which is suitable for use with a wide-range of abilities. Therefore, it is less susceptible to floor or ceiling effects. Receptive vocabulary is frequently used to match participants in studies examining theory of mind performance (e.g. O'Riordan *et al*, 1996; Zelazo *et al*, 1996). As the assessment battery was already fairly lengthy, it was decided to administer the short-form of the BPVS, in order to maximise co-operation from the participants and minimise inconvenience to families.

Theory of mind measures³.

Three theory of mind tasks drawn from the research literature were chosen to give to the research participants. First, participants were given the Smarties False Belief Task as described in Ozonoff *et al* (1991a). In this task the participant was shown a tube of Smarties and asked what they thought it contained. If the participant replied “Smarties”, the tube was then opened to show that it actually contained a pencil. The participant was then asked to predict what another person, who had never seen the tube, would think it contained. A pass was scored if they replied “Smarties”. This task is passed by the majority of normally developing three to four year old children (Perner *et al*, 1989).

Secondly, all participants were given a first order false belief task - the “Sally-Ann” task (Baron-Cohen *et al*, 1985). In this study the two protagonists in the story were called Roger and Billy. Participants were required to watch and listen to a short story in which Billy hid a marble, which was subsequently moved whilst he was not watching. The participant was then asked a false belief, a reality and a memory question, examples of which can be found in Appendix 1. Success on this task required the participant to know that Billy had not seen the marble being moved and therefore still believed that it was where he had hidden it. A pass was scored if they answered all three questions correctly. It has been found that normally developing children aged three to four pass this task (O’Riordan *et al*, 1996), but it is consistently failed by people with autism (Ozonoff *et al*, 1991).

Finally the participants were given a second order theory of mind task, which is a modification of the “Sally-Ann” task described above (O’Riordan *et al*, 1996). Second order theory of mind tasks require participants to predict one person’s thoughts about another person’s thoughts. The ability to make second order belief attributions of this kind develops later than mastery of first order false belief tasks. By the age of six to seven most children pass second order false belief tasks. (Perner & Wimmer, 1985).

³ Full text of these measures is given in Appendix 1.

Participants were required to watch and listen to a short story in which a marble was hidden by Billy and then moved by Roger. This time, unbeknown to Roger, Billy was watching Roger through a keyhole as the marble was moved. The participant was then asked a second order false belief question, a belief question, a reality question and a memory question, which are given in Appendix 1. A pass was scored if all four questions were answered correctly.

The theory of mind tasks selected, therefore, represented a range of measures which have been widely used in the research literature to examine social deficits. They covered a range of abilities, such that the simplest would be passed by normally developing three year olds, up to more sophisticated tasks which are not mastered in the normally developing child population until the age of seven. Therefore, these measures were appropriate for the developmental level of the participants in this study.

Executive function measures⁴

The measure chosen to investigate the executive function performance of the two groups was the modified Wisconsin Card Sorting Test (WCST-M) (Nelson, 1976). This task measures set maintenance skills, the ability to modify incorrect strategies and the ability to inhibit prepotent, but incorrect, responses (Ozonoff *et al*, 1991a). The WCST-M is a simplified version of the WCST (Heaton, 1981).

In the original WCST the participant is shown four key cards which show one red triangle, two green stars, three yellow crosses and four blue circles respectively. The dimensions along which the cards differ are colour, form and number. The participant is then given 128 other cards which contain all possible combinations of colour, form and number and asked to sort them out under the key cards. Correct performance requires the participant to sort the cards according to their colour, then their form and then their number. After each card is placed they are told whether it is correct or not. After ten unambiguous correct responses the rule for sorting is changed without the

⁴ Full details of the administration of the WCST-M are given in Appendix 1.

participant being told, and they must then try out different sorting strategies until finding the next correct category.

The WCST was modified to remove some of the ambiguities which arose from the original format of the test. Some of the cards in the original version share two or more attributes with a given key card, making it difficult to determine the sorting strategy being used by the participant. In addition, changing the sorting strategy without warning the participant can be stressful (Nelson, 1976). The WCST-M has all ambiguous cards removed (that is, cards to be sorted differ in only one attribute from the key cards). Removal of ambiguous cards leaves 48 to be sorted. The administration is also altered such that participants only have to make six correct responses before the sorting strategy is changed, and they are told that they should now use another rule for sorting the cards. The WCST-M is also considered to be less verbally mediated than the WCST and is a simpler and less ambiguous measure than the WCST (Nelson, 1976).

There are a number of dependent measures which can be calculated from the WCST-M. The total number of errors made and number of categories completed are measures of the participant's understanding of the nature of the categorisation of the task (Ozonoff *et al*, 1991a) and can be considered an assessment of a participant's overall proficiency at the task (Nelson, 1976). The number of perseverative errors made was also calculated.

Perseverative errors are made when a participant continues sorting cards according to a previously correct strategy. In the WCST-M any card which is sorted according to the same principle as an immediately preceding incorrect response is scored as perseverative (Nelson, 1976). More important than the absolute number of errors made is the proportion of them which are perseverative. This is the best predictor of frontal dysfunction derived from the WCST (Heaton, 1981). Therefore the percentage of perseverative errors was calculated.

The WCST-M is a simplified version of the WCST which itself has been extensively used to examine executive function deficits in children with autism (Pennington & Ozonoff, 1996). It has been used successfully with children as young as six (Chelune & Baer, 1986), and although derived down from a test designed for adults has been shown to be sensitive to developmental changes in executive functioning (Chelune & Baer, 1986).

Ethical approval⁵

Ethical approval for the study was obtained from the Local Research Ethics Committee. Families from Turk's sample (Turk, 1995) were then contacted by letter with details of the research and asked if they would be agreeable to participate in the study. Written consent was obtained from parents or guardians and the boys involved in the study. The families were then contacted by telephone and a mutually convenient time arranged to visit and carry out the assessment battery.

Ethical approval to approach the school was obtained from the Deputy Director of Education at Wandsworth Borough Council. Parents of boys identified as being suitable for the study were then approached by letter and sent information and consent forms. All the boys in the comparison group were tested at school at times convenient to them and their teachers.

Procedure

Boys with fragile X syndrome were seen at home or at school at times convenient to them and their families. The assessment battery was administered in the same order to all participants. The WISC-III^{uk} was administered first. After it was realised that participants were scoring close to the floor of this test, the short-form BPVS was added and administered directly after the WISC-III^{uk}. The three theory of mind measures were administered next in the order: Smarties, first order false belief task, second order false belief task. Finally the participants were given the WCST-M.

⁵ See Appendix 2 for letter sent to parents, information sheet and consent form.

During the assessment the participants were reminded that they could take breaks whenever they wanted. Administration of the battery took between 90 minutes and two hours. After the assessment the parents were given verbal feedback on their son's performance and, if requested, a written summary of the assessment.

The comparison group were assessed after the boys with fragile X syndrome. All participants in the comparison group were assessed at school in a separate, quiet classroom. The procedure was as given above. After the assessment parents were again given verbal feedback on their son's performance or a written report if they requested one.

Results

All the results are analysed for differences between groups. Following convention, the significance level for each analysis is set at 5%.

Sample characteristics

The sample consisted of two groups: eight boys with fragile X syndrome and eight with learning disabilities of unknown aetiology. Their age, Verbal IQ, Performance IQ, Full Scale IQ and BPVS raw score and mental ages are summarised in Table 1. BPVS raw scores and mental ages are presented for six fragile X boys: of the three boys with fragile X syndrome assessed prior to the addition of the BPVS to the test battery only one could be contacted to administer the BPVS. WISC-III^{uk} data are presented for seven fragile X boys: one did not complete the WISC-III^{uk}, but completed all other parts of the assessment battery.

Table 1
Descriptive characteristics of the sample

	Fragile X Group mean (SD) range	Comparison Group mean (SD) range
Age (months)	150.7 (16.3) 123-170	164.75 (9.6) 147-177
BPVS raw score	14.0 (3.0) 11-19	15.9 (3.1) 10-20
BPVS mental age (months)	76.0 (18.1) 58 - 107	83.9 (18.3) 53 - 114
Full Scale IQ	45.3 (3.2) 40-49	49.0 (7.2) 40-59
Performance IQ	49.7 (3.9) 46-56	55.6 (9.3) 46-71
Verbal IQ	48.1 (3.4) 46-55	49.4 (4.4) 45-57

Independent samples t-tests were used to compare the mean scores of these variables. All but the Full Scale IQ mean score had equal variances. Therefore the mean scores between groups were compared assuming equal variances for all dependent measures apart from Full Scale IQ for which equal variances were not assumed. The groups were not different in age [$t(14) = -2.097$, $p > .05$] or BPVS raw score [$t(12) = -1.142$, $p > .05$]. They did not differ in their mental age as assessed by the BPVS [$t(12) = -.800$, $p > .05$]. The groups were matched on Full Scale IQ [$t(9.9) = -1.316$, $p > .05$], Performance IQ [$t(13) = -1.566$, $p > .05$] and Verbal IQ [$t(13) = -0.600$, $p > .05$].

As the boys with fragile X syndrome had been studied before, some information was available from their previous assessments. The mean IQ of this group at Time 1 (ascertained from the BAS) and Time 2 (Full Scale IQ score from the WISC-III^{uk}) is given in Table 2.

Table 2

Comparison of previous IQ scores of the boys with fragile X syndrome
with those found in the current study

	Time 1 mean (SD)	Time 2 mean (SD)
IQ	66.9 (11.5)	45.3 (3.2)

The mean IQ scores from Time 1 and Time 2 were compared using a paired t-test. There was a highly significant difference between the means of the two groups [$t(6) = -5.288$, $p=.002$] and a moderate but not significant correlation between their IQ scores ($r=.507$, $p=.246$).

Theory of Mind tasks

The performance of the two groups on the three theory of mind tasks was examined by performing chi-squared tests, using Fisher's Exact Test, on the numbers in each group passing and failing each task. These results are summarised in Table 3.

Table 3

Proportion of each group passing each theory of mind task

	Fragile X Group Pass:fail	Comparison Group Pass:fail
Smarties	3:5	8:0
First Order False Belief	2:6	6:2
Second Order False Belief	2:6	5:3

Significantly more boys with fragile X syndrome failed the Smarties Task than boys in the comparison group [$\chi^2 (1, N=16) = 4.655$, $p=0.026$]. The difference between the two groups on the first order theory of mind task was not significant [$\chi^2 (1, N=16)$

=2.250, $p=0.132$]. There was no significant difference in the proportions of the two groups passing the second order theory of mind task [$\chi^2(1, N=16) = 0.000, p=1.000$].

The individual data for the first and second order false belief tasks were inspected to see whether those who failed the tasks had passed the control questions. For the fragile X group, five boys who failed the first order false belief task passed all the control questions. In the comparison group, one boy who failed the first order false belief task passed all the control questions. For the second order false belief task, three boys with fragile X syndrome who failed the task passed the control questions. In the comparison group, one boy who failed the task passed the control questions.

The above analyses were rerun with those who had failed a control question excluded. The proportion of boys with fragile X syndrome failing each false belief task was not significantly greater than the proportion of boys from the comparison group for both the first order [$\chi^2(1(N=14)) = 2.625, p=0.103$] and second order [$\chi^2(1(N=12)) = 0.000, p=1.000$] false belief tasks.

To examine whether passing the theory of mind tasks was a function of cognitive ability rather than group membership, the sample was divided into those who passed and failed each task regardless of their group membership. Those who failed control questions are excluded from this analysis. The mean score of each group was compared using independent samples t-tests to see if they differed on BPVS raw score, Performance IQ, Verbal IQ or Full Scale IQ.

For the Smarties Task, the two groups were significantly different on BPVS raw score [$t(12) = -2.274, p=.042$] but not significantly different on Verbal IQ [$t(13) = -.029, p>.05$], Performance IQ [$t(13) = -1.962, p>.05$] or Full Scale IQ [$t(13) = -1.587, p>.05$]. For the first order false belief task, those passing and failing did not differ on BPVS raw score [$t(10) = .828, p>.05$], Verbal IQ [$t(11) = -.456, p>.05$], Performance IQ [$t(11) = -1.496, p>.05$] or Full Scale IQ [$t(11) = -1.261, p>.05$]. Similarly, for the second order false belief task those passing and failing did not differ significantly on

BPVS raw score [$t(10) = .148, p > .05$], Verbal IQ [$t(11) = -.915, p > .05$], Performance IQ [$t(11) = .712, p > .05$] or Full Scale IQ [$t(11) = .923, p > .05$].

The fragile X group were then ranked in order of decreasing BPVS raw scores: two of those who passed the Smarties Task had the highest two BPVS scores. No BPVS data were available on the third boy with fragile X syndrome who passed the Smarties Task. The two boys with fragile X syndrome who passed the first and second order theory of mind tasks had the second and third highest BPVS scores in the group. Thus for the Smarties Task alone, passing or failing the task seemed to be related to BPVS raw score.

The mean BPVS mental ages of those passing and failing each theory of mind task is given in Table 4.

Table 4
Mean mental age (in months) of participants passing and failing
each theory of mind task

	Passing (SD)	Failing (SD)
Smarties	85.8 (17.8)	67.3 (10.9)
First Order False Belief	76.0 (25.4)	90.5 (25.4)
Second Order False Belief	80.7 (22.9)	81.0 (12.4)

Executive function tasks

Four variables were calculated from the WCST: the number of perseverative errors made; the total number of errors made; the total number of categories completed and the percentage of perseverative errors made. These results are shown in Table 5. One

participant from the fragile X group failed to complete any categories and his results have been excluded from these analyses.

Table 5
The mean perseverative and total errors of each group

	Fragile X Group (SD)	Comparison Group (SD)
Perseverative Errors	20.3 (12.1)	16.9 (15.5)
% Perseverative Errors	58.3 (28.8)	47.0 (34.4)
Total Errors	33.1 (4.7)	31.3 (7.6)
Number of Categories Completed	1.29 (.49)	1.13 (.35)

These means were compared using independent samples t-tests. The variance in each of these variables was equal between the two groups. There were no significant group differences between the mean number of perseverative errors made [$t(13) = .469$, $p > .05$] or percentage of perseverative errors made [$t(13) = .683$, $p > .05$]. The two groups did not differ in the mean number of total errors made [$t(13) = .566$, $p > .05$].

The number of categories completed by each group was calculated and the distribution across groups compared using a χ^2 statistic. The groups did not differ in the distribution of number of categories completed [$\chi^2(1, N=13) = .603$, $p = .446$].

The data from the two groups was collapsed and analysed for correlations between number of perseverative errors and total number of errors made and the overall cognitive ability variables using the Pearson product-moment correlation statistic. There were no significant correlations between percentage of perseverative errors

made and BPVS raw score ($r=.135$, $p>.05$), Full Scale IQ ($r=-.158$, $p>.05$), Performance IQ ($r=-.302$, $p>.05$) or Verbal IQ ($r=.232$, $p>.05$). The total number of errors made was not significantly correlated with BPVS raw score ($r=.239$, $p>.05$), Full Scale IQ ($r=-.217$, $p>.05$), Performance IQ ($r=-.330$, $p>.05$) or Verbal IQ ($r=.177$, $p>.05$).

The mean BPVS raw scores and mental ages of those completing zero, one or two categories on the WCST-M are given in Table 6.

Table 6
Mean BPVS raw scores of those completing different numbers
of categories on the WCST-M

Number of categories complete	Mean BPVS raw score (SD)	Mean BPVS mental age (SD)
0 (n = 1)	12 (0)	64.0 (0)
1 (n =10)	15.1 (3.1)	79.9 (17.6)
2 (n = 3)	16.0 (3.6)	88.0 (21.9)

Inspection of this data suggests that there is a clear developmental progression in the number of categories completed by participants from both groups.

Discussion

Participant characteristics

In total, sixteen boys fulfilled the criteria for inclusion in this study. Of the eight boys with fragile X syndrome, six had been diagnosed fragile X positive using the older, chromosome test for fragile X. Two had been tested using the newer and more sensitive DNA test. The DNA test is necessary to identify molecular variations in the expression of the fragile X chromosome (such as length of CGG expansion and degree of methylation of the CpG island). It has been shown that certain aspects of the

behavioural phenotype may be specific to subgroups which are homogeneous with respect to these molecular variations in the genotype (Wright-Talamante *et al*, 1996).

DNA testing was beyond the remit of the study, which allows for the possibility that the fragile X group are heterogeneous at the molecular level. This could compromise the finding of any group difference. However, with a relatively small sample size matching on a molecular level could produce very small subgroups. Furthermore the boys with fragile X syndrome had been evaluated by Turk (1995) as being typical developmentally and behaviourally of the larger cohort of boys with fragile X syndrome as ascertained by the newer DNA test.

The eight participants in the comparison group were recruited from a local, Inner London school. Thus there is a possibility of a difference between the two groups arising from geographical trends. The screening of the comparison group for autism was also less rigorous than that for the fragile X group, as it relied on inspection of the students' Statements of Special Educational Need. However, if any of the sample had an autistic spectrum disorder this would have added a conservative bias to the results, as they would be expected to show deficits in their theory of mind and executive function performance (Ozonoff *et al*, 1991a).

Furthermore, there is the possibility that some of the comparison group had fragile X syndrome. Again, any student with a known cause of their learning disabilities was excluded from the study, but the majority had not received any genetic testing to ascertain the cause of their learning disabilities. It was known that two of the comparison group had received negative fragile X tests. So, the comparison group could have contained students who were fragile X positive, or had another discrete genetic cause for their learning disabilities. Again, any students in the comparison group with fragile X would have introduced a conservative bias to the study, as they would have lessened any group differences.

Finally, in research with groups with specific aetiologies for their learning disabilities ideally two comparison groups are used: one of children with learning disabilities of

unknown aetiology to control for non-specific effects of growing up with learning disabilities, the other of participants with a different discrete genetic cause of their learning disabilities. This group can be used to investigate inter-syndrome comparisons (Turk, 1995). This study used a comparison group of boys with learning disabilities of unknown aetiology only, for the following reasons. First, recruitment of sufficient participants with a different discrete cause of their learning disabilities would have been very difficult due to time and resource constraints. Secondly, had a third group been added the numbers in each group required to have significant power in the analyses would have increased. Finally, the project had to be completed within a given time-frame, and addition of significant other participants would have made this unlikely.

Therefore it was decided that the study design would be a two group comparison only. This is sufficient to test that a given cognitive profile is specific to the fragile X group, and not a consequence of growing up with learning disabilities. However, the profile may be shared with another aetiologically-specific syndrome, so any mean differences found cannot be unequivocally attributed to a fragile X specific behavioural phenotype.

Group matching variables

The two groups were first matched for overall cognitive ability on the WISC-III^{uk}. There was no significant difference between the two groups on Verbal IQ, Performance IQ or Full Scale IQ. The variance in Full Scale IQ scores between the two groups was not equal. Inspection of the raw data showed that the comparison group had a much wider range of Full Scale IQ scores than the fragile X group, that is the fragile X group were more similar on this measure. The second measure used to match the groups was the BPVS. On this measure there was no difference in variance between the two groups, and they did not differ in their mean score. Although the means of the groups did not differ on any of the variables from the WISC-III^{uk} or the BPVS, the difference in variance between groups on the WISC-III^{uk} questions the validity of using the Full Scale IQ score as a variable on which to match the two groups.

The very low functioning of the fragile X group meant that many scored at floor on the WISC-III^{uk}. This instrument provides scaled scores which go down to a six year two month level of ability, yet some boys were scoring below that level on the individual subtests. This compromises the validity of using the dependent variables derived from the WISC-III^{uk} as measures on which to match the two groups. The BPVS is an instrument which covers a wider range of abilities (Dunn *et al*, 1982) and so is less prone to floor and ceiling effects. It is designed as a measure of receptive language ability (Dunn *et al*, 1982). There is some evidence that receptive language skills are a strength in the fragile X cognitive profile relative to their overall mental age (Freund & Reiss, 1991; Marans, Paul & Leckman, 1987), and so this measure could overestimate the ability of the fragile X group.

In aetiologically discrete groups, where syndrome-specific cognitive strengths and weaknesses are suspected, there is a real issue of which variables should be used to match groups. This relates back to the issue of discriminant validity (Bennetto & Pennington, 1996). Specific impairments cannot be inferred unless participants are shown to be relatively unimpaired in other measures which control for the general task requirements of the measure used. Therefore, it is proposed that the BPVS raw score provides the most reliable measure on which the two groups were matched, as it is not compromised by floor effects unlike the WISC-III^{uk}. However as it may overestimate the ability of the fragile X group compared to the comparison group, any group differences found on the experimental variables must be shown to have discriminant validity.

The final question to be addressed is the extremely low functioning of the fragile X group. It was hypothesised that the fragile X group would have mild to borderline learning disabilities. Their mean IQ score from Turk's 1995 study was 66.9. Instead, their abilities were in the mild to moderate learning disabilities range with a mean IQ score of 45. Although it is known that there is a gradual decline in IQ of boys with fragile X syndrome (Bennetto & Pennington, 1996) it is of the order of five to 14 IQ points, not the dramatic drop of nearly 22 IQ points seen here. It is hypothesised, therefore, that the large drop in IQ from previous testing is an artefact of the different

measures used on different testing occasions. Had the original comparison group been available for retesting this hypothesis could have been tested as the same longitudinal data were available for each group. In addition, if there had been access to other psychometric testing (such as that carried out for Statements of Special Educational Needs) then the developmental course of their IQ functioning could have been analysed. However, neither of these sources were available.

Evidence for theory of mind deficits

The three theory of mind measures administered were the Smarties Task (Ozonoff *et al*, 1991a; Perner *et al*, 1987) and a modified version of the first and second order Sally-Ann false-belief tasks (O'Riordan *et al*, 1996). The Smarties and first order Sally-Ann task are passed by the majority of normally developing three to four year old children (Perner *et al*, 1987). The ability to pass the second order false belief task develops later, with normally developing children passing it at around seven years of age.

There was a significant difference found between the two groups on their performance on the Smarties Task. Five out of eight participants with fragile X syndrome failed this task, compared to none of the comparison group. However, this cannot be attributed to a syndrome-specific impairment in this task. First, when the results were analysed in terms of mean BPVS scores of participants passing and failing this task, it was found that boys who passed the task had significantly higher BPVS scores than those who failed it. In addition, two of the boys with fragile X syndrome who passed this task had the highest BPVS scores in the fragile X group. The third boy with fragile X syndrome had not been tested using the BPVS, but inspection of his WISC-III^{uk} scores indicated that he had not scored at floor on either the Verbal or Performance subscales of this test (Verbal IQ = 50, Performance IQ = 52), and as such was almost one standard deviation above the mean of the fragile X group on this measure. From this evidence it can be assumed that his overall cognitive ability was likely to be above the average of the fragile X group.

Therefore, the difference between the two groups on this measure can be attributed to overall developmental level, rather than a syndrome-specific impairment in the fragile X group. Boys with fragile X syndrome with greater cognitive abilities were able to pass this test.

There were no significant differences in the proportions of each group passing the first and second order theory of mind tasks, although the difference in the case of the first order theory of mind task was just short of being significant. Again, more participants with fragile X syndrome failed this task. Only two boys with fragile X syndrome passed this task and they had the second and third highest BPVS raw scores in the fragile X group. There was no consistent relationship between BPVS raw score and participants passing and failing the first and second order theory of mind tasks. Indeed, the BPVS raw score of those failing the false belief tasks was higher than those passing it, although the variance is also much larger. Inspection of the data indicates that the mean BPVS score of those failing the false belief tasks was inflated by the participant with the highest BPVS raw score (20) failing this task.

The control questions asked with the first and second order false belief tasks show whether the task is failed due to a deficit in theory of mind abilities, or some other reason such as failing to remember the detail of the stories. For the first order false belief task, five of the six boys with fragile X syndrome and one of the boys in the comparison group passed all the control questions, but failed the task. For the second order false belief story, three boys with fragile X syndrome and one from the comparison group passed all the control questions but failed the false belief question. Analysis of the participants who failed only the false belief question showed that there was no significant difference in the proportions of boys with fragile X syndrome and learning disabilities of unknown aetiology passing each theory of mind task.

Therefore there is no evidence for any difference in theory of mind performance between boys with fragile X syndrome and boys with learning disabilities of unknown aetiology, although a proportion of each group failed the first and second order false belief tasks and passed the control questions. All the boys in this study had mental

ages (as assessed by the BPVS) above four years, the age at which typically developing children pass first order theory of mind tasks. The question arises, then, as to whether these results indicate wide-spread theory of mind deficits in children with learning disabilities regardless of aetiology. Alternatively, it can be argued that the BPVS does not give a reliable measure of overall cognitive ability.

There was a significant difference in the BPVS raw scores between those passing and failing the Smarties Task, but this difference was not significant for the other theory of mind measures. This pattern of results is consistent with a developmental progression in difficulty of theory of mind tasks from the Smarties test being the easiest, first order theory of mind more complex and second order theory of mind most difficult as shown by the increasing proportions of fragile X and control participants failing each task.

Evidence for executive function deficits

The executive function measures calculated were number and percentage of perseverative errors, total number of errors made and number of categories completed by each participant.

The WCST-M, therefore, provided three dependent measures. It has been argued that these can be divided into variables which measure executive function (the percentage of perseverative errors) and those which do not (number of categories completed and total number of errors: Ozonoff *et al*, 1991a; Nelson, 1976). Number of categories and total number of errors made are therefore measures of overall proficiency at the task (Nelson, 1976).

Chelune & Baer (1986) provide normative data on the WCST of children aged six to 12. The WCST is the more complex forerunner of the WCST-M. For children aged six to eight there is a developmental progression in number of categories completed and percentage of perseverative errors made (Chelune & Baer, 1986), as these variables have significant correlations with measures of overall cognitive ability. Therefore, for children of this developmental age range the two measures do not have

the same discriminant validity as with older age groups as performance on the task is mediated by overall ability. In adult populations there are very small correlations between overall cognitive ability as measured by the WAIS and number of categories completed ($r = .22$) and WAIS and percentage of perseverative errors made ($r = -.31$: Heaton, 1981)

In this study there was no difference between the fragile X and comparison groups on number of categories completed, total number of errors made or percentage of perseverative errors. The data were then collapsed across the two groups to examine the relationship between the WCST-M dependent variables and measures of overall ability. The BPVS scores of those completing different numbers of categories was calculated and showed a slight, but non-significant, increase in mean BPVS score as more categories were completed. There was no significant relationship between the total number of errors made and any of the measures of overall ability.

This pattern of results is consistent with the groups being at floor or at ceiling on the WCST-M. Inspection of the data leaves no doubt that both groups were performing at floor on this test. Their results can be compared, with caution, to the developmental norms provided by Chelune & Baer (1986). Chelune & Baer used the WCST with their sample. This is a more complex version of the WCST-M, and hence any performance above than the norms given may be due to the relative simplicity of the WCST-M. By the same token, any performance on the WCST-M below the norms represents a real deficit in performance on that measure.

Both groups in this study were significantly impaired on number of categories completed, total errors made and number of perseverative errors when compared to the norms for the youngest age group (six years old). The mean number of categories completed in Chelune & Baer's study was 2.73, it was 1.29 and 1.13 in the fragile X and comparison groups respectively. The mean number of perseverative errors made in Chelune & Baer's sample was 40.64 (out of a total number of cards sorted of 128) compared to a mean of 20.3 errors and 16.9 errors (out of 48 cards) in the fragile X and comparison groups respectively. Chelune & Baer do not give figures for the

number of total errors made, so the percentage of perseverative errors made cannot be compared. It is clear, however, that both groups in the current study perform well below the levels of the youngest age group in the Chelune & Baer sample.

Therefore it can be concluded that dependent measures derived from the WCST-M are not sensitive measures of executive function in this population. Neither are they sensitive to overall levels of cognitive functioning, as shown by the lack of correlation between dependent variables from the WCST-M and BPVS raw scores.

Suitability of measures

The discussion above has examined the evidence from this study for a difference in performance on theory of mind and executive function measures between a group of boys with fragile X syndrome and a group of boys with learning disabilities of unknown aetiology. No group differences were found between any of the dependent variables derived from the measures used which could be attributed to group membership rather than overall level of cognitive ability.

The question arises, therefore, as to whether this lack of difference is a reliable finding leading to the conclusion that the two groups do not differ in these cognitive abilities, or whether the lack of difference can be attributed to another cause.

First, as discussed above, there were a number of factors in the recruitment of the participants for this study which could have lessened the likelihood of finding a difference in performance between the two groups. The fragile X group was likely to contain boys who were heterogeneous at the molecular level of their genotype. It has been suggested that aspects of the behavioural phenotype may be specific to certain forms of the genotype (Wright-Talamante *et al*, 1996). This study was designed to investigate the cognitive processes which might underlie the abnormal social behaviour seen in boys with fragile X syndrome. The lack of homogeneity of this research group is particularly salient, therefore, if it is suspected that abnormalities in social interaction are limited to a subset of all boys with fragile X syndrome.

The available evidence suggests that, when children with fragile X syndrome and autism are excluded from samples, “autistic-like” behaviours which are considered as part of the fragile X syndrome behavioural phenotype are not consistent amongst boys with fragile X syndrome. For example, Turk & Graham (1997) found that 20% of their fragile X syndrome sample had some abnormality of eye contact (17.1% avoided eye contact, 2.9% had some other abnormality of eye contact). The proportion of boys with learning disabilities of unknown aetiology or Down’s Syndrome with abnormal eye contact was significantly lower, with 4.2% and 5.0% respectively showing eye contact avoidance. Therefore, abnormality of eye contact is not found in all boys with fragile X syndrome but is significantly more common than in boys with learning disabilities of unknown aetiology or with Down’s Syndrome.

Unfortunately, Turk & Graham’s study did not have DNA genetic data for all their participants with fragile X syndrome so it can not be ascertained whether abnormalities of eye gaze are specific to certain forms of the genotype. An equally plausible alternative explanation is that it shows an acceptable degree of constancy to be considered as part of the fragile X syndrome behavioural phenotype. The lack of a finding of abnormality of gaze in the remainder of the fragile X syndrome group can be attributed to other, non-specific variations in the genotype and specific environmental influences of the participants in the group (Bennetto & Pennington, 1996).

There is insufficient evidence, therefore, to ascertain whether the possibility of lack of molecular homogeneity in the fragile X syndrome group would mask any group differences in the measures used. More significant is the possibility that children in the comparison group could have had autism, an autistic spectrum disorder, or fragile X syndrome. Due to project constraints, the comparison group were screened for autism and fragile X syndrome on the basis of information in their Statements of Educational Special Need and the knowledge of their teachers. It was known that two of the comparison group (25%) had had negative fragile X tests, but no genetic data were available for the other students. All the comparison group seemed socially appropriate during testing, and no “autistic-like” abnormalities of social interaction were reported

in their school records. Obviously this does not constitute as rigorous a screening for autism as that in the fragile X syndrome group, and so it has to be concluded that there is a possibility that undiagnosed autism or fragile X syndrome in the comparison group could have reduced any between group differences found.

Secondly, the results obtained on the theory of mind and executive function measures suggest that the measures used were too complex for the developmental age of most of the participants. The lowest functioning failed the most simple theory of mind task, and both groups scored more poorly on the WCST-M dependent variables than typically developing six year olds. There were no correlations between the WCST-M variables and measures of overall ability, as is commonly found, suggesting that the WCST-M is not a sensitive and appropriate measure for these groups.

The failure of participants on the Smarties and first order false belief task is somewhat surprising. These tasks are passed by normally developing three to four year old children (Perner *et al*, 1987). As assessed by the BPVS, the lowest mental age in this study was four years five months, with the average six years eight months. Therefore all the participants could have been expected to pass these tasks. Two possibilities for this pattern of results are that the BPVS inflated the verbal mental age of participants i.e. it is not a valid measure of mental age in these groups, or that theory of mind deficits are widespread in children with learning disabilities regardless of aetiology.

In both the fragile X syndrome and comparison groups there were children who passed the Smarties and first order theory of mind task as expected. In the fragile X syndrome group, but not the comparison group, a significant number failed the Smarties Task. There was a very clear trend of this being related to BPVS raw score. The pattern was less clear for the first order theory of mind task. Two control students failed this task whose BPVS scores were relatively high. A high proportion of fragile X syndrome participants failed this task, but the relationship to BPVS raw score was not as consistent as for their performance on the Smarties Task.

It is suggested that this pattern of results is consistent with the BPVS providing overestimates of the general ability of the fragile X syndrome group. It has been proposed that children with fragile X syndrome show a syndrome-specific profile of better verbal than performance abilities (Freund & Reiss, 1991) and that they have a characteristic weakness in sequential processing (Dykens *et al*, 1987). The BPVS is a task requiring single word processing which may be a relative skill in boys with fragile X syndrome (Freund & Reiss, 1991). Therefore, a measure of global ability based on this task is likely to overestimate their abilities if assessed using a measure of a range of cognitive abilities, and hence lead to their failure on tasks which seem to be within their capabilities.

The BPVS may have inflated the scores of the fragile X group relative to the comparison group, as described above. However, failure on the first and second order theory of mind tasks cannot be wholly explained by the use of the BPVS to match groups. A proportion of both groups failed first and second order false belief tasks but passed the control questions, undermining the argument that failure is due to overall cognitive impairment.

Therefore it cannot be concluded that the lack of difference between the two groups studied is reliable. The method by which the comparison group was recruited leaves open the possibility that students in this group had either an autistic spectrum disorder or fragile X syndrome. The measure used to match groups, whilst covering an appropriate range of abilities for use with these groups, may have had the effect of inflating the fragile X syndrome scores. This may explain the widespread failure of fragile X syndrome participants on theory of mind tasks. Had more simple measures of theory of mind and executive function been used, this question could have been more validly addressed.

These measures were chosen based on an estimate of the cognitive level of the fragile X syndrome group which was significantly higher than their actual abilities. Had the group been in the predicted range of abilities then many of these difficulties would not have arisen. The groups could have been matched on a wider range of variables than

was possible, given that many of the fragile X syndrome group scored below the floor of the WISC-III^{uk}, rendering dependent variables derived from it invalid.

Clinical implications

One of the reasons for studying behavioural phenotypes is that there is an increasing body of work which suggests that clinical and educational interventions may need to be tailored to the strengths and needs of particular groups of children with learning disabilities (Sobesky, 1996). Thus, if significant discrepancies had been found between boys with fragile X syndrome and those with learning disabilities of unknown cause on measures of executive function or theory of mind this could have informed interventions with this group.

The initial findings suggest that boys with fragile X syndrome are more similar to boys with learning disabilities of unknown aetiology on measures of theory of mind and executive function than they are different. Failure on measures of theory of mind are not significantly more prevalent in the fragile X group than the comparison group. The two groups did not show any significant differences in their executive function performance as measured by the WCST-M. From this study there was no evidence for a syndrome-specific profile of abilities on these measures.

What may be more important from a clinical perspective is the poor performance in general on theory of mind and executive function measures in both groups. Their mean performance on each dependent measure from the WCST-M was far below that expected from a typically developing six year old child. This suggests that one of the features of learning disabilities in general might be deficits in executive function performance. This suggests that children with learning disabilities may need more help with tasks which are hypothesised to require executive function, such as structuring information, problem solving and reasoning.

This study also highlights the importance of assessing a range of cognitive abilities when assessing children with learning disabilities, as measures may not have the same concurrent validity in this population as in normally developing children.

Future research

This study does not exclude executive function or theory of mind deficits from being important in the development of the abnormalities in social behaviour seen in fragile X syndrome. What may be important is to research a developmental perspective on their acquisition. In a clinical setting, where a significant number of children with developmental disabilities are referred, it may be appropriate to assess each with a standardised battery of measures, to begin to obtain a true developmental perspective on patterns of behavioural and cognitive development for different causes of learning disabilities. For example, the Vineland Adaptive Behaviour Scales (Sparrow, Balla & Cicchetti, 1984) is a normative, standardised assessment instrument which can be used to assess domains of personal and social functioning in infants. This could be usefully used to identify patterns of functioning in different syndromes.

Were the study to be repeated, simpler measures would be required. Given the difficulty outlined above in using the WISC-III^{uk} and BPVS to match groups, a simpler measure of overall ability would be employed. In the case of syndromes such as fragile X syndrome, where syndrome-specific styles of processing have been proposed (Dykens *et al*, 1987) it may be appropriate to match groups using measures based on cognitive processes, such as the Kaufman Assessment Battery for Children (K-ABC: Kaufman & Kaufman, 1983) or the BAS (Elliot *et al*, 1982).

Alternatively the same tasks could be employed with higher functioning children with fragile X syndrome, such as individuals with the pre-mutation. However, the finding of a difference in one form of the genotype cannot be taken as evidence for an impairment in all genotypic expressions of the syndrome.

Finally, this study highlights the importance of using a comparison group of children with learning disabilities when looking for specific impairments in groups of children with learning disabilities of a known aetiology. Comparisons groups of mental age matched normally developing children are not suitable. This study suggests that all children with learning disabilities may perform poorly on certain measures compared to typically developing children.

Conclusion

This study aimed to compare the theory of mind and executive function performance of boys with fragile X syndrome and boys with learning disabilities of unknown aetiology. The theory of mind measures ranged in complexity from those which are passed by the majority of typically developing three year olds to tasks requiring the ability of a typically developing seven year old. The measure of executive function used had been shown to be appropriate for use with typically developing six year old children, and should therefore have been within the range of abilities of these participants.

On these measures there was no difference in performance between boys with fragile X syndrome and boys with learning disabilities of unknown aetiology. However, this may not be a reliable result. The data suggest that the groups were performing at floor on the measures used, and the lack of difference between the groups can therefore be attributed to the measures being unsuitable for the developmental age of the participants in this study, rather than reliably reflecting equal performance in both groups.

The poor performance of the participants in this study is somewhat surprising, and it is concluded that it arose from a number of factors. First, the fragile X syndrome group were predicted to have far higher overall abilities than proved to be the case. Had they been in the range expected, the measures would have been developmentally appropriate. The discrepancy between their predicted and actual abilities could not have been predicted from the literature, as the magnitude of discrepancy was much greater than that found in studies of boys with fragile X syndrome. It is therefore

hypothesised that the discrepancy is due to the different assessment measures used, in addition to any actual decrease in ability.

However, even allowing for the lower overall ability of the boys with fragile X syndrome, their poor performance on the theory of mind measures used was unexpected. Most of the fragile X syndrome group failed the Smarties Task - the simplest theory of mind task used. It has been argued that this does not represent a reliable syndrome-specific impairment in theory of mind in boys with fragile X syndrome compared to those with learning disabilities of unknown aetiology. Inspection of the data showed that boys with fragile X syndrome who failed the Smarties Task had lower BPVS raw scores than those who passed, suggesting that level of receptive ability (which is related to overall cognitive ability: Dunn *et al*, 1982) was a more important factor in completion of the task than group membership.

The performance of both groups on the first and second order false belief stories suggest that deficits on theory of mind tasks are no more prevalent in the fragile X group than the comparison group. At this level of overall ability there may not be a syndrome-specific impairment in theory of mind: this does not preclude specific impairments in simpler tests of precursors of theory of mind.

Finally it is concluded that, in investigating syndromes which cause developmental disabilities, the issue of discriminant validity is of paramount importance. Where people with a given syndromes have an cognitive profile which is uneven (such as is the case with fragile X syndrome: Dykens *et al*, 1994) care must be taken to account for this and ensure that groups are matched on relevant task variables. In this case, the BPVS may have provided an overestimate of the abilities of the fragile X syndrome group.

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Appendix 1

Protocols for administering the theory of mind tasks and WCST-M

Procedure for theory of mind tasks

Smarties task

Child is shown a tube of Smarties and asked: "What do you think is in here?" [Answer: "Smarties."]. The lid is taken off and they are shown a pencil inside. The lid is replaced and they are asked; "What did you think was in the tube?. What would [friend's name] think was in the tube?"

A pass is scored if the child replies, "Smarties"; the task is failed if they reply "a pencil".

First order false belief story

The first story is about Roger and Billy. This is Roger and this is Billy.

Prompt question: Which doll is Roger? Correct? Yes No

Prompt question: Which doll is Billy? Correct? Yes No

Billy puts the marble in his basket. Then he leaves the room. While he is out Roger takes the marble out of the basket and puts it in his box. Then he goes out of the room.

False belief question:

When Billy comes back in, where will he look for the marble?

Reality question:

Where is the marble really?

Memory question

Where was the marble in the beginning?

Second order false belief story

Now this is a different story.

This is Billy and this is Roger again.

Prompt question: Which doll is Roger? Correct? Yes No

Prompt question: Which doll is Billy?

Correct?

Yes No

Billy puts the marble in his basket. Then he leaves the room.

While he is out Roger takes the marble out of the basket and puts it in his box, but Billy is watching him do this through the keyhole.

False belief question:

When Billy comes back in, where does Roger think Billy will look for the marble?

Belief question:

Where will Billy look for the marble?

Reality question:

Where is the marble really?

Memory question

Where was the marble in the beginning?

Instructions for completing the modified WCST

Lay out the four key cards in front of the participant. Then say:

“Here we have four key cards. I want you to sort these cards (*indicate the response cards*) under the key cards according to certain rules: but the whole point of the test is that I shall not tell you what the rule is. I want you to find out by trying out different rules and each time I shall tell you whether it’s right or wrong. Now go ahead and try to find out the rule.”

After 6 consecutive correct responses say:

“The rules have now changed. I want you to find another rule.”

Appendix 2

Letter of introduction, information sheet and consent form sent to participants

Letter of introduction sent to parents of fragile X participants and children with learning disabilities of unknown aetiology from Turk's 1995 study.

Mr. & Mrs. Smith
1 The High Street
Bromley
Kent

31 January 1997

Dear Mr. & Mrs. Smith,

I am in my final year of training to be a Clinical Psychologist. At the moment I am working in the Child Mental Health Learning Disability Service with Dr. Jeremy Turk. I am interested in social understanding in children with learning disabilities. I have obtained your name from Dr. Turk, and understand that you agreed with him that you would be willing to be approached about possibly participating in further studies.

As part of my time working with Dr. Turk's team I hope to undertake some research examining the social behaviour of children with learning disabilities. This research is being undertaken in collaboration with Dr. Turk, Consultant Child Psychiatrist and Dr. Maria Callias, Consultant Clinical Psychologist. The study has been approved by St. George's Hospital Ethics Committee.

The study uses tests to examine social understanding in children and examine whether or not the child appreciates that other people act according to their beliefs, rather than those of the child. Other tasks assess abilities like planning and flexibility of thinking. Most children find these tests fun and enjoyable to do.

I hope that you will be willing for your son, John, to take part in this research. I have included an information sheet with this letter which gives details of the study. If you

have questions about any aspect of it, please contact me at St. George's Hospital to discuss them.

Whether or not you are willing to take part in this research, I would be grateful if you and John could complete a consent form and personal information sheet and return it to me in the envelope enclosed. Please keep one copy of the consent form for your information. I will then contact you to arrange a convenient time for me to visit you and work through the tests with John. After the tests are completed I will be able to give you a verbal report of the pattern of John's abilities. Once the study is complete I will send you a summary of the results.

I hope that you find the enclosed information interesting, and that you and John will want to participate in this research. If you have any questions or concerns about this research please don't hesitate to contact me to discuss them.

Many thanks for your time in considering this information.

Yours sincerely

Claire Garner
Clinical Psychologist in Training

Dr. Jeremy Turk
Consultant Child Psychiatrist

Dr. Maria Callias
Consultant Clinical Psychologist

A comparison study of boys with fragile X syndrome and boys with learning disabilities on their ability to think flexibly and understand social behaviour

Information for parents and participants

This study is designed to examine two particular areas of thinking and reasoning - the ability to think flexibly and solve problems, and the ability to understand social behaviour.

Why is this important? Well, by identifying what different groups are good at, and what they might need more help with, we can begin to design specific ways to help groups of people manage their particular problems.

What happens if I agree to take part? I (Claire Garner) will contact you and arrange a convenient time when I can visit your son at home (or at school if that would be better). Your son would complete a number of tasks designed to assess the areas mentioned above, and a general measure of a wide range of aspects of thinking. These tests would take between 1½ and 2 hours to complete. During this time, your son can take breaks whenever he wants. These tests are often used in clinical and research work and are not unpleasant to do. In fact, many people find that they are like games. All the tests can be done in one visit, after which you will not be asked to do anything more.

If you have any questions, or would like any more information before making your decision to participate, please get in touch with Claire Garner, at the number given on the cover sheet. Please return the consent form in the envelope provided. If you do not want to take part, that's fine. You are free to withdraw from the study at any time without giving a reason. Your decision to participate (or not) will **not** affect your future care.

Signed by the person in charge of the project: _____ Date: _____

The District Medical Ethical Committee has approved the above statement:

Signed by the Chairman of that Committee: _____ Date: _____

A comparison study between boys with fragile X syndrome and with learning disabilities on their ability to think flexibly and understand social behaviour

Consent form

Please note: this form should be read and signed by the participant and his parent or guardian. Please keep a copy of this form for your information.

I confirm that I have read the information sheet and have had the chance to ask questions about the study. Any questions which I have raised have been answered. I am satisfied that I have received enough information about the study.

I understand that my participation in this research is voluntary. I can drop out of the study at any time and don't have to give a reason. If I decide that I don't want to participate, it won't affect my future care.

Therefore I agree to take part in this study YES NO
(Please cross out as applicable)

Signed _____ Date: _____

(NAME IN BLOCK LETTERS) _____

I am agreeable to my son taking part in this study

Signed _____ Date: _____

(NAME IN BLOCK LETTERS) _____