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Gene-Environment Interactions and the Functional Analysis of Challenging Behaviour in Children with Intellectual and Developmental Disabilities

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This thesis is dedicated to the memory of DB who taught me why all of this matters so much

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Abstract

Challenging behaviour displayed by people with intellectual and developmental disabilities represents a socially significant problem that severely restricts the quality of life of those who display such behaviours and their families. To date the study and conceptualisation of such behaviours has been characterised by genetic and environmental determinism. It seems likely that gene-environment interactions (GxE) may play a critical role in challenging behaviour displayed by some people with intellectual and developmental disabilities. The current thesis provides a conceptual and empirical examination of GxE as applied to challenging behaviour. The two empirical studies presented in the current thesis aimed to examine the hypothesis that genetic events may function as a type of motivating operation for challenging behaviour. In study one, parents of children with FXS, SMS and of a mixed etiology group of children with intellectual and developmental disabilities were interviewed about the function served by their child's challenging behaviour using the *Questions About Behavioral Function* (QABF; Matson & Vollmer, 1995) scale. Both within- and between-group differences were found. Children with FXS were less likely to display attention-maintained challenging behaviours; children with SMS were more likely to display pain-related challenging behaviours. In study two, experimental functional analysis methods were used to examine this question in a group of eight children with FXS and six children with SMS. No child with FXS displayed an attention-maintained response class of challenging behaviour. In contrast four children with SMS displayed a response class of challenging behaviour that was at least in part attention-maintained. The findings are discussed in light of the research literature on challenging behaviour in FXS and SMS. The findings are related to GxE and it is suggested that a developmental systems model, consistent with the principles of radical behaviourism, may help to move the field beyond the limitations of genetic and environmental determinism.

Chapter I. Challenging Behaviour. Epidemiology, Impact and Explanatory Models

"Selection is not a metaphor, model, or concept; it is a fact. Arrange a particular kind of consequence, and behaviour changes. Introduce new consequences, and new behaviour will appear and survive or disappear."

Skinner (1984, p. 503)

Chapter Overview

In the current chapter, challenging behaviour displayed by some people with intellectual and developmental disabilities is established as a socially significant behaviour. Such behaviours are relatively prevalent in this population and, in the absence of effective intervention, appear to be characterised by their chronicity. Challenging behaviour has a pervasive negative impact on the quality of life of both those who display it and others who share their environment. Given the negative consequences of challenging behaviour across the lifespan, it is argued that further research is required to further current understanding of the factors involved in the evocation and maintenance of such behaviour.

A number of psychological models have been used to account for challenging behaviour in people with intellectual and developmental disabilities. Particular emphasis is given to applied behaviour analysis and the behavioural phenotypic approach. The relative contributions of each approach are discussed. Functional analysis is the hallmark of the applied behaviour analytic approach to challenging behaviour and assumes that such behaviours are influenced by their environmental antecedents and consequences. The behavioural phenotype approach has been characterised by the identification of associations between particular genetic syndromes, such as fragile X syndrome and Smith-Magenis syndrome, and various topographies of challenging behaviour. It is argued that each of these two approaches have, with few exceptions, developed in isolation of the other, resulting in an apparent dichotomy between 'nature' and 'nurture' in the study of challenging behaviour.

The developmental systems model, which is drawn on throughout the current thesis, is introduced as a means of bridging this divide. An overview of the remainder of the thesis is provided.

Challenging behaviour, as displayed by some people with intellectual and developmental disabilities, exerts a serious impact; both on the lives of those who display it and on those individuals who share their environment. The physical and social impact of such behaviours demands a systematic response that renders challenging behaviour *inefficient*, *ineffective* and *irrelevant* (Horner & Day, 1991). To this end the current thesis aims to further explore the interaction between environmental and genetic factors in the development and subsequent maintenance of challenging behaviour.

In this chapter challenging behaviour is defined as a socially significant problem. Explanatory models that have been used to account for why some people with intellectual and developmental disabilities display challenging behaviour are discussed and particular emphasis is given to applied behaviour analysis, as well as the behavioural phenotype approach. Finally the aims of the current thesis are discussed and justification provided for each subsequent chapter.

Challenging Behaviour. Definitions, Epidemiology and Impact.

As this work is being conducted within a field awash with labels terms that are used throughout are now defined.

Definitions

Intellectual and developmental disability.

The term intellectual and developmental disability is used in preference to other labels, such as mental retardation or learning disability, as it avoids confusion with terms that have acquired disparaging connotations or those that have different meanings in different countries (Emerson, 2001). Intellectual and developmental disability is best understood in terms of functional impairments that result from historic organism-environment interactions (Bijou, 1966). The American Association on Intellectual and

Developmental Disability provide the following definition of intellectual and developmental disability:

> *A disability characterized by significant limitations both in intellectual functioning and in adaptive behavior as expressed in conceptual, social, and practical adaptive skills. This disability originates before the age of 18.* (Luckasson et al., 2002, p. 3)

Challenging behaviour.

Terms such as 'abnormal' and 'problematic' are often applied to behaviours displayed by individuals with intellectual and developmental disabilities. Such terms imply that the problem lies within the person. The term challenging behaviour was originally adopted in North America by The Association for Severe Handicaps (TASH) to place an emphasis on the environmental determinants of such behaviours, shifting the focus away from the individual as the locus of the problem. Indeed the term suggests that, "*if services could rise to the challenge then they would cease to be challenges*" (Blunden & Allen, 1987, p. 14).

Emerson (1995) provides a definition of challenging behaviour, which emphasises the inter-play between the topography and consequences of the behaviour as well as the social context in which it occurs:

> *culturally abnormal behaviour(s) of such an intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to ordinary facilities (p. 3).*

Challenging behaviour is a label that has been applied to a wide range of behaviours, all of which may vary in their form, frequency, severity and aetiology. It may include aggression, property destruction, self-injurious behaviour, stereotypy, pica, non-compliance, screaming, and sexualised behaviour. Those behaviours that occur frequently and/or result in serious environmental disruption, such as aggression, are especially likely to be defined as challenging (Lowe & Felce, 1995).

Challenging behaviour is socially defined and whether a behaviour is labelled challenging is dependent on the context in which that behaviour occurs (Hastings & Remington, 1994a, 1994b; Wolfensberger, 1972). The capacity of a service to prevent, detect, support and manage challenging behaviour will also influence the extent to which that behaviour is regarded 'challenging' (Mansell, McGill, & Emerson, 1994; Quereshi, 1994). In addition, the application of such a label, may also be reinforced by some of the consequences that follow its use, such as access to services or funding (McGill, Clare, & Murphy, 1996).

Epidemiology of Challenging Behaviour

Estimates of both the incidence and the prevalence of challenging behaviour have varied, due in part to the difficulty in reaching a consensus over definition. There has been considerable variability between epidemiological studies with regards to the forms of behaviour studied, the size and characteristics of samples and the data collection methods employed (Rojahn & Esbensen, 2002).

A small number of studies have examined the overall prevalence of challenging behaviour within the total population of a given geographical area. Such studies suggest that between 5 - 19% of people with intellectual and developmental disabilities display challenging behaviour (Borthwick-Duffy, 1994; Emerson & Bromley, 1995; Emerson et al., 2001; Joyce, Ditchfield, & Harris, 2001; Kiernan & Quereshi, 1993; Lowe et al., 2007; Quereshi & Alborz, 1992). For example, Lowe and colleagues (2007) report that some 10% of individuals with an intellectual disability in an area of Wales displayed challenging behaviours rated as serious.

More commonly, studies have restricted sampling to specific sub-populations such as children attending special schools (Kiernan & Kiernan, 1994) or have examined the prevalence of specific topographies or classes of behaviour, such as self-injurious behaviour (Cooper, Smiley, Allan et al., 2009; Maurice & Trudel, 1982; Oliver, Murphy, & Corbett, 1987; Schroeder, Schroeder, Smith, & Dalldorf, 1978) and aggression (Cooper, Smiley, Jackson et al., 2009; Harris & Russell, 1989).

There has been a paucity of research to address the incidence of challenging behaviour. Two recent studies by Cooper and colleagues reported on the incidence of aggression and self-injury in an area of Scotland and reported a two year incidence of 1.8% and .6% respectively (Cooper, Smiley, Allan et al., 2009; Cooper, Smiley, Jackson et al., 2009). Although, findings on this have been somewhat equivocal (Schroeder et al., 1978), it does appear that once established in an individual's repertoire and in the absence of effective intervention, challenging behaviour may be characterised by its chronicity (Green, O'Reilly, Itchonen, & Sigafoos, 2005; Murphy et al., 2005).

Impact of Challenging Behaviour

Challenging behaviour can be damaging to the physical and psychological well-being of the individual and those around them. For example, repeated self-injury may lead to the development of calcified haematomas, sensory loss, brain-damage and may increase the risk of mortality (Nissen & Haveman, 1997). Although this relationship is bi-directional, challenging behaviour is a major cause of stress and anxiety for care staff and the family (Hastings, 2002a, 2002b; Jenkins, Rose, & Lovell, 1997). Environmental restriction and abusive practices are unfortunately a relatively common response to those whose behaviour challenges services (Rusch, Hall, & Griffin, 1986; Saloviita, 2002).

Many 'treatments' for challenging behaviour can exert a deleterious impact on the quality of life of individuals who display such behaviours. A number of studies have demonstrated that the use of typical anti-psychotic medication represents the most prevalent treatment for people who display challenging behaviour (e.g., Robertson, Emerson, Pinkney, Caesar et al., 2005). Not only are such drugs associated with a range of negative side-effects but they are often ineffective in treating challenging behaviour (e.g., Tyrer et al., 2008)[1]. Physical interventions appear to be over-used as a response to challenging behaviour, particularly in specialist services for people with intellectual and developmental disabilities who display challenging behaviour (Robertson, Emerson, Pinkney, Caesar et al., 2005). Such physical interventions are associated with pain and distress (Hawkins, Allen, & Jenkins, 2005; Sequeira & Halstead, 2001). Finally, many people with intellectual and developmental disabilities are denied access to evidence-based interventions for challenging behaviour, such as applied behaviour analysis (Emerson et al., 2000; Robertson et al., 2000). When behavioural interventions have been applied there has been a historical tendency for them to be aversive in nature (Scotti, Evans, Meyer, & Walker, 1991).

The social consequences of challenging behaviour can be as debilitating as the sometimes more apparent physical consequences. People with intellectual and developmental disabilities and challenging behaviour are likely to be offered limited opportunities to make choices (Robertson, Emerson, Hatton et al., 2001; Shaddock et al., 1993), have small and restricted social networks (Robertson, Emerson, Gregory et al., 2001), have limited access to community amenities and facilities (Hill & Bruininks, 1984) and tend not to be included in the community (Robertson, Emerson, Pinkney, Caeser et al., 2005). Such individuals are also likely to spend large amounts of time disengaged, receiving minimal contact from care staff (Felce, Lowe, & Blackman, 1995; Mansell, 1995). In addition, people with intellectual and developmental disability who

[1] The authors of a Cochrane review into the use of neuroleptic medication for the management of challenging behaviour concluded that: "*It is debatable whether the use of antipsychotic medication for certain people with intellectual disabilities and challenging behaviour is ethical outside of a randomised controlled trial.*" (Brylewski & Duggan, 1999, p. 369).

display challenging behaviour are often at a heightened risk of placement breakdown (Allen, 1999; Mansell et al., 1994).

Together, these findings demonstrate that challenging behaviour is a socially significant problem, one that exerts a pervasive and enduring negative influence upon the individual's quality of life. Despite the array of negative consequences, challenging behaviour remains characterised by both its prevalence and by its chronicity. An important task for researchers and clinicians alike is to account for this apparent paradox.

Understanding Challenging Behaviour: Explanatory Models

Various approaches have attempted to explain the development and maintenance of challenging behaviour. Typically such approaches have reflected the wider divisions that lie within psychology as a science and as such have been relatively incompatible (Lee, 1992). There have, however, been some attempts to provide an integrated model of challenging behaviour by incorporating variables at the biological, cognitive, environmental and wider social level (see Gardner, Cole, Davidson, & Karan, 1986; Murphy, 1997)[2]. Given that such frameworks have been the exception rather than the rule, however, the following discussion is placed in the context of the radical behavioural critique of 'mentalistic' approaches to challenging behaviour.

Mentalistic Approaches to Challenging Behaviour

The dominant approach to psychology assumes that a person's behaviour is the result of the workings of internal structures, such as cognitions or beliefs, which enable

[2] The integration of cognitive variables is also possible within the developmental systems model. Whilst not of primary concern in the current thesis, the concept of the *'cognitive endophenotype'* has received considerable attention in recent years and it has been proposed that certain genetic syndromes may have a characteristic cognitive style (Woodcock, Oliver, & Humphreys, in press). See chapter six for further discussion of this issue.

the individual to represent and interpret their external world (e.g., Tolman, 1932). Environment is usually viewed as important only in the extent that it leads to the development of those structures that lie within the individual, which ultimately 'cause' behaviour (Chiesa, 1992; Lee, 1992).

Such models have been used by some to account for challenging behaviour displayed by people with intellectual and developmental disabilities. For example challenging behaviour has been described as an attempt to alleviate guilt (Cain, 1961), as a response to the uncomfortable physical sensation of thinking (Sinason, 1993) and as a means of defending the 'self' from a perceived threat (Jahoda, Trower, Pert, & Finn, 2001). Recent approaches to the treatment of aggression in people with intellectual and developmental disability have emphasised the role of emotional variables, such as anger, and intervention has focused on modifying the 'cognitive scripts' thought to play an important role in such behaviours (Novaco & Taylor, 2008; Taylor, Novaco, Gillmer, & Thorne, 2002).

One of the strongest challenges to such 'mentalistic' accounts comes from radical behaviourism (Skinner, 1953). According to radical behaviourism, mentalistic interpretations are based on a Cartesian division of the individual, in which the body and the mind are held as two separate entities. Skinner argued that such explanations are tautological, in that causality is inferred by reference to the impact of the inferred construct, such as a belief or cognition, on behaviour; which is then taken as evidence for the very existence of the same construct[3]. As Skinner states:

[3] This point was eloquently made by Woodworth (1921) who stated: "*Instead of 'memory', we should say 'remembering'; instead of 'thought' we should say 'thinking'; instead of 'sensation' we should say 'seeing, hearing', etc. But, like other branches, psychology is prone to transform its verbs into nouns. Then what happens. We forget that our nouns are merely substitutes for verbs, and go hunting for the things denoted by the nouns; but there are no such things, there are only the activities that we started with.*" (p.5-6).

The practice confuses the order of events and leads to unfinished causal accounts (Skinner, 1963, p. 278).

Radical behaviourism argues that mentalistic accounts of behaviour are unnecessary as a more parsimonious and complete explanation can be provided without reference to hypothetical variables that lie in the gap between stimulus and response (Skinner, 1953). Mentalistic accounts have also been critiqued on pragmatic grounds (Baum, 1994). If such variables were shown to be capable of predicting and influencing behaviour then they would warrant further analysis (R. Hawkins, 1992; Skinner, 1974). However, there is a lack of data to support the view that behavioural change can be brought about solely by the manipulation of cognitions or other such variables (Sturmey, 2006).

Applied Behaviour Analysis

Radical Behaviourism

Radical behaviourism is the philosophy that underpins the experimental and applied analysis of behaviour. Methodologically, Skinner established a means of conducting an experimental analysis of behaviour, characterised by the direct observation of changes in the response rate of a single organism over time, within well-specified experimental conditions (Kazdin, 1978). Conceptually, Skinner argued all human behaviour is orderly, developing and altering according to basic universal principles. Radical behaviourism abandons the Cartesian division of man, in favour of a holistic approach, in which the person is seen as an indivisible whole, dynamically interacting with his or her environment (Chiesa, 1992).

Radical behaviourism provides a 'relational' account of human behaviour in that it emphasises the identification and manipulation of functional relationships between behaviour and its environmental determinants. The scientific aim of radical behaviourism lies in the prediction and influence of behaviour (Craighead, 1981; Hayes, Hayes, & Reese, 1988). Therefore the description and explanation of complex patterns of behaviour is achieved by relating identifiable environmental events to behavioural principles whilst avoiding recourse to unobservable and inferred constructs.

Radical behaviourism argues that all forms of behaviour evolve through a history of interaction between the individual and their environment (Baer, Wolf, & Risley, 1968; Bijou & Baer, 1961). Contingencies between behaviour and its consequences and associated antecedents serve both to evoke and maintain such operant behaviour (Skinner, 1953).

Applied Behaviour Analysis

As general principles of behaviour, inductively developed through laboratory experimentation with animals, began to be applied to human behaviour (e.g., Fuller, 1949), there became a need for the development of an appropriate forum for such research, which ultimately led to the publication of the *Journal of Applied Behavior Analysis* in 1968. In their seminal paper Baer, Wolf and Risley (1968) established the cornerstone principles of applied behaviour analysis[4]:

1) Applied. Socially significant behaviours or events should be the primary focus of research.

2) Behavioural. Changes in behaviour are to be the primary dependent variable.

[4] Baer, Wolf & Risley (1968) described applied behaviour analysis as: "*the process of applying sometimes tentative principles of behavior to the improvement of specific behaviors, and simultaneously evaluating whether or not any changes noted are indeed attributable to the process of application- and if so, to what parts of that process. In short, analytic behavioral application is a self-examining, self-evaluating, discovery-orientated research procedure for studying behavior.*" (p.91).

3) Analytic. Studies should provide a 'believable demonstration' that behaviour is related to environmental events.

4) Technological. The techniques adopted to change behaviour should be operationalised.

5) Conceptually systematic. Procedures and their effects should be related to behavioural principles.

6) Effective. Should lead to changes in socially significant behaviour.

7) Generalisable. Should generalise over time, individuals and settings.

Functional Analysis

According to the applied behaviour analytic approach, therefore, challenging behaviour, like all behaviour, is a functional and adaptive response to contingencies that prevail in the individual's historic and immediate environment. Early behavioural approaches to challenging behaviour, however, were characterised by the application of powerful contingencies of reinforcement and punishment which aimed to override the contingencies responsible for the maintenance of such behaviours (Mace, 1994). This approach was commonly termed behaviour modification and involved the application of technology in the absence of any clear value-base (Emerson & McGill, 1989). Punishment-based interventions, such as time out and over-correction were consistently applied to eliminate challenging behaviour (Cataldo, 1989). However reinforcement-based techniques such as the differential reinforcement of other behaviour often resulted in mixed outcomes (Carr, Taylor, Carlson, & Robinson, 1989). The ineffective, and in some cases counter-therapeutic, outcomes of such procedures could have been predicted given that they were applied somewhat prescriptively and without due consideration of the contingencies underlying the development and maintenance of behaviour (Iwata, Vollmer, Zarcone, & Rodgers, 1993). Such practice was somewhat removed from the central aims of behaviour analysis (Mace, 1994). Furthermore the reliance on often dehumanising and coercive procedures such as punishment received considerable criticism and was the subject of great debate within (e.g., LaVigna & Donnellan, 1986; VanHouten et al., 1988) and outside (Guess, Helmstetter, Turnbull, & Knowlton, 1987; McGee, 1987) the behaviour analytic field.

Strategies for effective behaviour change require an understanding of the variables that underpin behaviour (Carr, 1977; Owens & Ashcroft, 1982). Towards this end, functional analysis has become the hallmark of applied behaviour analysis (Hanley, Iwata, & McCord, 2003). A functional analysis involves the identification of the variables that influence behaviour and the relationships that exist between those variables through experimental manipulation. The antecedents and consequences that evoke and maintain challenging behaviour are the focus of functional analysis and their manipulation is central to effective intervention.

The conceptual paper by Carr (1977) represented a watershed in the applied analysis of challenging behaviour. In this paper Carr argued that self-injurious behaviour could be understood in the same way as any other response (i.e., as an operant behaviour) and provided a number of hypotheses regarding the variables involved in the maintenance of such behaviours. This, and early empirical papers (Carr, Newsom, & Binkoff, 1976; Lovaas, Freitag, Gold, & Kassorla, 1965), acted as a stimulus for the development of a comprehensive functional analysis methodology first reported in a seminal study by Iwata et al. (1982/1994).

Using a multi-element design, Iwata et al exposed nine individuals with severe self-injurious behaviour to four conditions, designed to provide an 'analogue' of the relations originally described by Carr (1977). The study aimed to test whether self-injurious behaviour was maintained by either socially-mediated processes (i.e., positive reinforcement, negative reinforcement) or by its automatic consequences. Experimental conditions included *social disapproval*, in which self-injurious behaviour was followed by the contingent provision of 10-sec of attention; *social demand,* in which a difficult demand was removed for 30 seconds contingent on self-injurious behaviour; *alone*; in which the person was left alone in a room and no consequences were provided for the behaviour; and a control *play* condition in which the person received non-contingent access to toys and attention. Self-injury was shown to vary both between and within subjects, providing direct empirical evidence that self-injurious behaviour was a function of different socially and non-socially mediated sources of reinforcement. Later studies incorporated additional conditions, such as a *tangible* condition (Day, Horner, & O'Neill, 1994) or *social avoidance* condition (Hagopian, Wilson, & Wilder, 2001) to test for additional functions of challenging behaviour.

Functional analysis promoted a return to the foundations of behaviour analysis and has served to improve the link between basic and applied science (Mace, 1994). Not only has this approach allowed unique behaviour-environment relations to be uncovered but it has also encouraged a greater integration with basic behavioural science (Mace, Lalli, Lalli, & Shea, 1993). The recognition that challenging behaviour serves as a functional response has enhanced the social acceptability of assessment and intervention procedures (Donnellan, Mirenda, Messaros, & Fassbender, 1984). Indeed the advent of functional analysis has been associated with the adoption of more precise antecedent and reinforcement-based interventions and an apparent decrease in the use of punishment (Pelios, Morren, Tesch, & Axelrod, 1999).

Contingencies of Reinforcement

Functional analysis has stimulated increasingly refined and complex questions regarding the personal and environmental variables responsible for the maintenance of challenging behaviour (Carr, 1994). Challenging behaviour has consistently been found to be maintained by four general classes of reinforcement. Multiple studies across a number of different behavioural topographies have shown challenging behaviour may be maintained by socially or non-socially mediated forms of positive reinforcement and negative reinforcement. The following section briefly summarises research to have investigated each class of reinforcement as it relates to challenging behaviour displayed by people with intellectual and developmental disabilities. Although the conceptual utility of the distinction between positive- and negative reinforcement has been questioned (i.e., Baron, 2005; Michael, 1975)[5], the following discussion shall, for heuristic purposes, preserve the distinction.

Socially mediated positive reinforcement.

Challenging behaviour may be reinforced by the contingent presentation of socially mediated stimuli (Carr, 1977). When such a stimulus change leads to an increase in a given response this is termed positive reinforcement. The contingent provision of social contact has been found to function as a type of positive reinforcement for challenging behaviour. A number of descriptive studies have shown that attention is a relatively common consequence for challenging behaviour in natural settings (e.g., Emerson et al., 1996; McKerchar & Thompson, 2004; Thompson & Iwata, 2001). Descriptive methods are, however, ill suited to capturing functional relationships that exist between behaviour and its consequences (Iwata et al., 1993).

A more 'believable' demonstration of the role of attention has been provided by studies that involve the experimental manipulation of the antecedents and consequences of behaviour. Iwata et al (1982/1994) reported that the differentiated responding of four participants in their study provided direct evidence that self-injurious behaviour was either exclusively or partially maintained by social attention. This finding has been

[5] Michael (1975) has recommended that the descriptive terms of 'positive' and 'negative' be abandoned, arguing that it is illogical to describe a stimulus change in terms of the application of (positive) or removal of (negative) stimuli; indeed there are many situations in which it may be relatively unclear whether behaviour is reinforced by access to tangibles, such as food, or by the removal of an aversive state, such as hunger.

consistently replicated across a number of topographies of behaviour and forms of attention. An epidemiological review of such studies showed that of 536 individual data sets, 25.3% revealed patterns consistent with attention-maintained behaviour (Hanley et al., 2003). Recent studies have shown that the type of attention provided during such conditions can alter the probability of attention-maintained behaviour, suggesting that individuals vary as to the types of attention that function as reinforcers (e.g., Kodak, Northup, & Kelley, 2007). Even more conclusive have been those studies that have based an intervention on the results of such an analysis (Lovaas et al., 1965; Lovaas & Simmons, 1969; Vollmer, Iwata, Zarcone, Smith, & Mazaleski, 1993). For example, functional communication training (FCT) involves the differential reinforcement of an alternative, socially acceptable response that forms part of the same response class as challenging behaviour. Such interventions have been shown to be effective in reducing attention-maintained challenging behaviour (e.g., Carr & Durand, 1985; Durand, 1999; Kurtz et al., 2003).

Although easily confounded with social attention, challenging behaviour may also be positively reinforced by the contingent provision of preferred tangible activities or items (Moore, Mueller, Dubard, Roberts, & Sterling-Turner, 2002). Descriptive studies suggest that the provision of such tangibles may be a relatively common consequence for challenging behaviour in natural settings (McKerchar & Thompson, 2004; Thompson & Iwata, 2001). Experimental analyses have shown challenging behaviour may be maintained by the provision of tangibles (Day, Rea, Scussler, Larsen, & Johnson, 1988). Approximately 10.1% of functional analyses are consistent with positive reinforcement via the provision of tangibles (Hanley et al., 2003). Finally, a number of studies have shown that interventions, such as FCT, that involve the differential reinforcement of behaviours in the same response class as challenging behaviour can be effective in reducing tangible-maintained challenging behaviour (Day et al., 1988; Durand, 1999; Durand & Kishi, 1987; Hagopian et al., 2001).

Non-socially mediated positive reinforcement.

Some behaviours are positively reinforced by their automatic consequences. Unlike socially mediated stimuli, such types of reinforcement are not typically open to direct observation or manipulation and their presence must therefore be inferred (Hanley et al., 2003). In a functional analysis automatic positive reinforcement is inferred by the occurrence of behaviour in the absence of social stimuli or by heightened levels of responding across all social and non-social conditions (Iwata et al., 1982/1994). Epidemiological reviews of functional analyses reveal that between 15%-21% of individuals show patterns of responding consistent with an automatic reinforcement hypothesis (Derby et al., 1992; Hanley et al., 2003; Iwata et al., 1994). More detailed analyses have added informal components to further elaborate existing hypotheses and to rule out alternative explanations regarding the source of automatic reinforcement (Goh et al., 1995; Kennedy & Souza, 1995; Piazza et al., 1998; Piazza, Hanley, & Fisher, 1996). Interventions that interrupt the contingency between behaviour and its automatic consequences, such as response blocking, and those that involve providing 'matched' forms of stimulation provide greater evidence in support of the automatic reinforcement hypothesis (e.g., Kennedy & Souza, 1995; Rapp, Miltenberger, Galenky, Ellington, & Long, 1999).

Socially-mediated negative reinforcement.

In some instances challenging behaviour may be negatively reinforced by the removal or attenuation of socially-mediated aversive stimuli (Carr, 1977). Behaviour may be a function of negative reinforcement when the response is followed by a) the complete avoidance of aversive stimuli, b) a delay in the presentation of aversive stimuli,

c) the attenuation of the strength of aversive stimuli or d) the alleviation of an aversive condition (Skinner, 1969).

A number of descriptive studies (e.g., Edelson, Taubman, & Lovaas, 1983; Emerson et al., 1996) suggest that socially mediated negative reinforcement may be a relatively common consequence for challenging behaviour.

Studies that involve functional analysis methodology have demonstrated that challenging behaviour may increase when difficult tasks are presented and when escape from task-related demands is provided contingently (e.g., Carr, Newsom, & Binkoff, 1980; Mace & West, 1986). This finding has been replicated across a range of behavioural topographies. Hanley et al. (2003) found that 34.2% of functional analyses showed patterns consistent with a negative reinforcement hypothesis. Challenging behaviours may also be negatively reinforced by variables other than escape from demands, such as the removal of social attention (Hagopian et al., 2001) or social proximity (Oliver, Oxener, Hearn, & Hall, 2001). Interventions that have manipulated the contingency between challenging behaviour and escape (Richman, Wacker, Asmus, & Casey, 1998; Zarcone, Iwata, Vollmer, Smith, & Mazaleski, 1993) and those that have involved the differential reinforcement of socially acceptable behaviours within the same response class as negatively reinforced challenging behaviours (Day et al., 1994; Durand, 1999; Hagopian et al., 2001) have provided further evidence that challenging behaviour may be maintained by a process of negative reinforcement.

Non-socially-mediated negative reinforcement.

Challenging behaviour may also be negatively reinforced by its automatic consequences (Carr, 1977). For example it has been hypothesised that in some cases self-injurious behaviour may be maintained by negative reinforcement, via the relief or attenuation of aversive stimulation, such as pain (Carr, 1977; Lovaas, Newsom, & Hickman, 1987). In such cases, challenging behaviour may serve to automatically decrease levels of aversive stimulation (DeLissovoy, 1963). Evidence for the existence of such relations in relation to challenging behaviour is scarce. However, there is some evidence to suggest that self-restraint may for some individuals be maintained by the removal of the painful consequences associated with self-injury (Fisher, Grace, & Murphy, 1996; Silverman, Watanabe, Marshall, & Baer, 1984).

Summary

In sum, challenging behaviour has been hypothesised by some to be the external manifestation of internal psychological pathology (e.g., as in psychodynamic and cognitive formulations). Applied behaviour analysis represents an empirical, relational approach to the study of human behaviour and explicitly attends to the environmental determinants of such behaviour. A plethora of evidence exists to suggest that challenging behaviour serves as a functional and adaptive response for the individual. As Durand (1990) states:

> *Behavior problems...are reasonable behavioral adaptations necessitated by the abilities of our students and the limitations of their environments (p. 6).*

Functional analysis provides a model to guide not only the identification of the factors that influence challenging behaviour but also the relations between such factors. Treatment decisions that are based on a functional analysis are likely to be more effective, constructional and thereby socially valid, than those based on alternative rationales.

The Behavioural Phenotype

Crick and Watson's discovery of DNA in 1953 and the subsequent mapping of the human genome transformed the status of genes from the hypothetical to the directly observable. This development has had a dramatic influence on the field of intellectual and developmental disabilities and has ushered in a new wave of research based on the study of the 'behavioural phenotype'. Certain genetic syndromes associated with intellectual and developmental disability are characterised by the heightened prevalence of challenging behaviour. This suggests that genetic factors may play a role in the causation of challenging behaviour.

The Concept of the Behavioural Phenotype

The term behavioural phenotype refers to a broad array of developmental and behavioural characteristics associated with different genetic syndromes. Most pertinent to the current discussion is the observed association between behavioural problems and particular genetic syndromes. William Nyhan (1972) was the first to apply the term 'behavioural phenotype' to describe the self-mutilative behaviours characteristic of individuals with Lesch-Nyhan syndrome. However, the term also refers to motor, social, linguistic and cognitive abilities observed in individuals with biologically determined syndromes associated with intellectual and developmental disability.

A considerable amount of disagreement exists regarding the use of the term behavioural phenotype and much of this disagreement surrounds the specificity of the relationship between the genetic disorder and behaviour. Some have argued that the gene-behaviour relationship should be totally-specific (i.e., *unique* to the syndrome and *universal* to all people diagnosed with that syndrome) before being termed a behavioural phenotype (Berrini & Kahn, 1987; Flint & Yule, 1996)[6]. Others, such as Dykens (1995), have adopted a more probabilistic definition of the behavioural phenotype:

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[6] Berini and Kahn (1987) define a phenotype as being the "*observable or measurable expression of a gene or genes*". Likewise, Flint and Yule (1996) argue that: "*A behavioural phenotype should consist of a distinctive behaviour that occurs in almost every case of a genetic or chromosomal disorder, and rarely (if at all) in other conditions" (Flint & Yule, 1996, p. 69).*

A phenotype may best be described as the heightened probability or likelihood that people with a given syndrome will exhibit certain behavioural and developmental sequalae relative to those without the syndrome (Dykens, 1995, p. 523)

Whilst Hodapp and Dykens (2001) have noted that both environmental and genetic factors contribute to within-syndrome variability (and presumably consistency); there remains a degree of ambiguity regarding the extent to which such behaviours are considered genetically determined.

Evidence for the behavioural phenotypes associated with fragile X syndrome and Smith-Magenis syndrome is now reviewed to provide an example of the behavioural phenotype approach to challenging behaviour. Both these syndromes are the focus of the empirical work in this thesis.

Fragile X Syndrome and the Behavioural Phenotype

Fragile X syndrome (FXS) is the most common inherited cause of intellectual and developmental disability, with an estimated prevalence of 1 in 4,000 in males and 1 in 8,000 in females (Crawford et al., 1999; Turner, Webb, Wake, & Robinson, 1996). The genetic locus of FXS lies in a single gene on the X chromosome, known as the Fragile X Mental Retardation 1 *(FMR1)* gene located at Xq27.3 (Verkerk et al., 1991). FXS is caused by an increase in the number of trinucleotide repeats of cytosine-guanine-guanine (CGG) on the long arm of the X chromosome in the *FMR1* gene. A full mutation comprises of more than 200 repeats, leading to hypermethylation of the cytosines in the *FMR1* promoter region and thereby preventing the transcription of the *FMR1* gene into mRNA and the translation of the fragile X mental retardation protein (FMRP). Low levels of FMRP have been shown to be associated with poorer developmental outcome (e.g., Bailey, Hatton, Tassone, Skinner, & Taylor, 2001), although the size of this effect is not large, suggesting that other variables may also play an important role. A number of studies have shown that there are differences in both the brain structure and functioning of individuals with FXS when compared to controls (Gothelf et al., 2008; A. L. Reiss, Aylward, Freund, Joshi, & Bryan, 1991; A. L. Reiss, Patel, Kumar, & Freund, 1988; Rivera, Menon, White, Glaser, & Reiss, 2002).

FXS is associated with a number of physical characteristics; these include a long face, large ears, a prominent jaw and macroorchidism. FXS is also associated with a characteristic pattern of adaptive behaviours, with daily living skills for example representing a relative strength for individuals with FXS (e.g., Dykens et al., 1996). There also appears to be a characteristic pattern of speech in FXS, which is characterised by the use of repetitive speech and tangential language (Belser & Sudhalter, 2001; Sudhalter & Belser, 2001).

Challenging behaviour.

Several studies have investigated the association between FXS and challenging behaviour. Hessl et al (2002) reported that boys and girls with FXS (aged 6-17 years) scored higher on the Child Behavior Checklist (*CBCL*; Achenbach, 1991) than control siblings without FXS. Likewise, Hatton et al (2002) using the CBCL reported that 49% of their sample of young boys with FXS (aged 4-12 years) scored within the clinical or borderline range for total problem behaviour. A controlled comparison study conducted by Steinhausen et al (2002) using the Developmental Behavior Checklist (DBC; Einfeld & Tonge, 1995) reported that children with FXS (aged 5-16 years) showed higher levels of total problem behaviours than children with Prader-Willi syndrome or Tuberosis Sclerosis but lower overall levels than children with Fetal Alcohol Syndrome. These studies suggest that children with FXS, especially males, may be particularly likely to display challenging behaviour but provide little information regarding the topography of these behaviours.

Autism and hyperactivity.

Several studies have investigated the association between FXS and autism. It appears that a relatively high proportion of children with FXS either meet the diagnostic criteria for autism or score highly on autism rating scales. Several studies have used rating scales, such as the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1988) to investigate the prevalence of autism in individuals with FXS (e.g., Bailey, Hatton, Skinner, & Mesibov, 2001; Bailey et al., 1998; Demark, Feldman, & Holden, 2003; Hatton et al., 2006). Other investigators (Budimirovic et al., 2006; Clifford et al., 2007; S. Hall, Lightbody, & Reiss, 2008) have relied on autism diagnostic tools such as the Autism Diagnostic Observation Schedule (ADOS-G; Lord et al., 2000). Hall et al (2008) reported that some 50% of males and 20% of females with FXS (aged 5-20 years) met the diagnostic criteria for autism using the ADOS-G. Children with both FXS and autism appear to have a particularly poor developmental outcome in comparison to those with FXS alone (Bailey, Hatton, Skinner et al., 2001). Other studies, however, have failed to replicate the association between FXS and autism (Einfeld, Molony, & Hall, 1989). Indeed, Einfeld et al suggest that the high levels of gaze avoidance and hand flapping exhibited by children with FXS may lead to a misdiagnosis of autism.

Boys with FXS are also likely to present with attention deficits and/or hyperactivity (Baumgardner, Reiss, Freund, & Abrams, 1995; Sullivan et al., 2006; Turk, 1998). In a controlled study, Sullivan, Hatton et al (2006) reported that there was a higher prevalence of Attention Deficit Hyperactivity Disorder (ADHD) in children with FXS (aged 7-13 years) than in mental age matched controls. Likewise, Baumgardner, Reiss, Freund and Adams (1995) found 73% of their sample of 31 males with FXS met the diagnostic criteria for ADHD.

Gaze aversion and social anxiety.

Gaze aversion appears to be particularly characteristic of both boys and girls with FXS (Cohen, Vietze, Sudhalter, Jenkins, & Brown, 1989; Hall, DeBernadis, & Reiss, 2006; Meerenstein et al., 1996). Meerenstein et al (1996) report that 80-88% of males with FXS exhibit poor eye contact. Cohen, Vietze, Sudhalter, Jenkins and Brown (1989) had children interact for 10 minutes with a stranger and 10 minutes with their mother, and reported that young males with FXS were more likely to engage in escape behaviours (turning body away, running away, looking away from task, not looking at the adult) than were individuals with Down syndrome, autism, and typically developing controls. A recent study by Hall, Maynes and Reiss (2009) demonstrated that a percentile shaping procedure, in which successively greater durations of eye contact were differentially reinforced, was effective in reducing gaze aversion in three boys with FXS, suggesting that such behaviours, although phenotypic, are open to environmental influence.

Self-injurious behaviour.

A relatively large proportion of individuals with FXS display self-injurious behaviours. Hall, Lightbody and Reiss (2008), using the Self-Injury Checklist (SIB-C; Bodfish et al., 1995) reported that self-injury occurred in 58% of boys and 17% of girls with FXS. Likewise, Symons et al (2003), using a bespoke self-injury questionnaire investigated self-injurious behaviours displayed by boys with FXS (aged 1-12 years) and reported that 58% of participants had displayed self-injurious behaviour at some point during their lifetime, 81% of whom continued to self-injure. As part of a larger study Hessl et al (2008) reported the results of the Behavior Problems Inventory (BPI; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001) for a sample of 50 males with FXS (aged 8-24 years). Some 79% of the sample was reported to have displayed some form of self-injurious behaviour over the preceding two months. The average frequency of these behaviours was reported to be weekly.

Some studies have provided a relatively fine-grained analysis of those topographies of self-injurious behaviour associated with FXS. In their study, Hessl et al (2008) reported that the most common topographies of self-injury were self-hitting (50% of the sample) and self-biting (30%). Meerenstein and colleagues (1996) reported that 63.9% of males with FXS in their sample (aged 3 months-60 years) displayed hand-biting. Hall, Lightbody and Reiss (2008) reported that 45.2% of males and 6.9% of females in the sample displayed self-biting, 16.2% of males and 0% females displayed self-hitting and 22.6% of males and 13.8% of females displayed rubbing or scratching. In their study Symons et al (2003) reported that the most commonly reported form of self-injurious behaviour in boys with FXS was hand- and finger-biting (72%), followed by head hitting (41%) and picking/pulling of the skin and hair (34%). Other reported behaviours included skin rubbing/scratching, head banging, insertion of fingers/objects into body cavities and leg hitting. Over 50% of children displayed multiple forms of self-injurious behaviour. Self-injurious behaviours were most commonly directed to the head (63%), front and back of the hands (50-59%) and the front of the arms (41%). Symons et al reported a correlation between FMRP levels and both the age of onset and location of self-injury. FMRP levels did not, however, differ between those boys with FXS who displayed self-injurious behaviour and those who did not.

Stereotypical behaviours.

It appears that individuals with FXS may also be more likely to display some topographies of stereotypical behaviour. For example, Hessl et al (2008) reported that 98% of their sample had displayed stereotypical behaviours over the preceding two months, with the most commonly reported topographies including repetitive hand movements (50% of the sample), waving or shaking arms (48%), and waving hands

(44%). The average frequency of stereotypical behaviour reported in this sample was daily. Meerenstein (1996) reported that 83.8% of males with FXS engaged in hand-flapping. In a recent large-scale comparison study, Moss Oliver, Arron, Burbidge and Berg (2009) used the Repetitive Behaviour Questionnaire to examine the phenomenology of repetitive behaviours in individuals with FXS, Angelman syndrome, Cornelia De Lange syndrome, Cri du Chat syndrome, Prader-Willi syndrome, Lowe syndrome, Smith-Magenis syndrome and a control group of individuals with intellectual disability of heterogenous causes. In comparison to other groups, individuals with FXS (aged 6–47 years) were more likely to engage in hand stereotypy, lining up of objects, echolalia, and to have restricted conversation and a preference for routine.

Aggressive behaviours.

Very few studies have examined the occurrence of aggressive behaviours in FXS. Hessl et al (2001) compared problem behaviours in boys and girls with FXS using the *CBCL*, which has a specific subscale to measure aggressive behaviours. Whilst over 12% of boys and girls with FXS scored in the clinically significant range for aggressive behaviours there were no significant differences between children with FXS and their siblings on this subscale. Hatton and colleagues (2002) reported that 17% of children in their sample displayed aggressive behaviour that fell in the borderline or clinically significant range on the CBCL. Merenstein and colleagues (1996) reported that 56.3% of males with FXS in their sample displayed aggressive behaviour and 31.9% had been reported to have had a violent outburst.

Using the BPI, Hessl et al (2008) reported that some 75% of the sample had displayed aggressive behaviours at some point over the preceding two months, with hitting others (49% of the sample) and kicking others (30%) the most commonly reported topographies. The average frequency of aggressive behaviours was reported to be weekly.

Smith-Magenis Syndrome and the Behavioural Phenotype

Smith-Magenis syndrome (SMS), first described in two infant males by Smith et al (1982), is caused by an interstitial deletion of chromosome 17p11.2. Several candidate genes have been identified in the deleted region of chromosome 17p11.2 (Elsea et al., 1997) suggesting that SMS may be a contiguous genetic syndrome (Greenberg et al., 1991). However, recent research suggests that mutations in *RAI1 (Retinoic Acid Induced Gene)* are specifically associated with most features of the SMS phenotype in individuals who did not have a detectable 17p11.2 deletion (Slager et al., 2003) and haploinsufficiency of the RAI1 gene is now thought to be the primary cause of the disorder (Edelman et al., 2007). SMS has a reported prevalence of 1/25,000, although this is likely to be an underestimation. Most cases of SMS are sporadic and there is an equal sex ratio (Greenberg et al., 1991).

SMS is associated with a number of physical abnormalities. In a phenomenological study involving 27 individuals, Greenberg et al (1996) described some of the physical characteristics associated with the syndrome. Common physical features include characteristic facial features (mid-face hypoplasia, prominent forehead, epicanthal folds, broad nasal bridge, down turned mouth with cupid's bow, ocular and ear anomalies), short stature, hoarse and deep voice, speech delay, signs of peripheral neuropathy (including decreased sensitivity to pain and temperature, reduced leg muscle mass, gait disturbance, and muscle weakness), high incidence of myopia (with and without retinal detachment). Less common findings include congenital heart defects, seizures, facial clefts and urinary tract anomalies. In addition, intelligence quotient scores range between 20 and 78, with most lying in the moderate range of intellectual disability (Greenberg et al., 1996). There are relatively few reports of the cognitive abilities of individuals with SMS. At least three studies have reported that daily living skills remain an area of relative weakness in comparison to other areas of adaptive behaviours and what would be expected given the relatively high intellectual functioning of people with SMS (Greenberg et al., 1996; Madduri et al., 2006; Udwin, Webber, & Horn, 2001).

Smith, Dykens and Greenberg (1998) reported a high level of sleep disturbance in a sample of 39 individuals with SMS (aged 1-32 years); these difficulties included problems falling asleep, frequent night-time awakenings and excessive daytime sleepiness. In a recent meta analysis Edelman et al (2007) reported that 88% of a sample of 50 individuals with SMS had some form of sleep disturbance. The sleep disturbance found in SMS has been associated with a disturbance in the secretion of melatonin (De Leersnyder et al., 2001). Some evidence exists to support the use of night time melatonin alongside beta-blockers (to reduce daytime elevation of melatonin) in the treatment of sleep disturbances in SMS (De Leersnyder et al., 2007).

Challenging behaviour.

Initial case reports of SMS made reference to a number of stereotypical, self-injurious and aggressive behaviours displayed by individuals with the syndrome (e.g., Colley, Leversha, Voullaire, & Rogers, 1990; Finucane, Konar, Haasgivler, Kurtz, & Scott, 1994; Lockwood et al., 1988; Smith et al., 1986; Stratton et al., 1986). For example, Smith et al (1986) noted the presence of self-injurious behaviours in 6 of the 9 cases of SMS. These early case reports suggested that self-squeezing, hand biting, self-pinching, picking at sores, hitting the head or body, and tearing or picking finger/toenails or the skin around the nails and inserting objects into bodily orifices all occurred in people with SMS.

The results of subsequent studies suggest that such behaviours may form part of the behavioural phenotype for SMS.

Between-group comparison studies suggest that individuals with SMS display challenging behaviours of greater severity than that generally seen in other groups. For example, Clarke and Boer (1998) found that individuals with SMS (aged 5-32 years) had higher scores across all domains of the *Aberrant Behavior Checklist* (ABC; Aman, Singh, Stewart, & Field, 1985) than did individuals with Cri du Chat syndrome or Prader-Willi syndrome. Likewise, Dykens and Smith (1998) reported that children with SMS had higher total scores on the *CBCL* than did children with Prader-Willi syndrome or a mixed control group of children with intellectual disability.

Self-injurious behaviour.

A number of studies have investigated the phenomenology of self-injury associated with SMS (Dykens, Finucane, & Gayley, 1997; Dykens & Smith, 1998; Finucane, Dirrigl, & Simon, 2001; Greenberg et al., 1996; Martin, Wolters, & Smith, 2006); typically these studies have been hampered by relatively low numbers of participants.

Estimates of the prevalence of self-injurious behaviour in SMS vary notably between studies. Greenberg et al (1996), for example, report 67% of their sample of 27 individuals with SMS (aged 1-30 years) as displaying some form of self-injurious behaviour. Martin et al (2006) report that 78.9% of their sample of 19 children with SMS (aged 2-12 years) displayed self-injury, Dykens and Smith (1998) report that 92.2% of their sample of 35 individuals with SMS (aged 4-20 years) displayed at least one form of self-injury. Similarly Finucane et al (2001) report 96.6% of their sample of 29 individuals with SMS (aged 1-49 years) as displaying some form of self-injury. Sloneem (2005) recently reported a study involving some 32 people with SMS, using both the Challenging Behaviour Checklist (Harris, Humphreys, & Thomson, 1994) and the Challenging Behaviour Interview (Oliver et al., 2003), in which 96.9% of the sample (aged 6-39 years) displayed self-injurious behaviour. In a recent meta-analysis Edelman et al (2007) reported that 89.5% of individuals with SMS were reported to have displayed self-injurious behaviours. It appears that the prevalence of self-injury in

SMS is greater than that typically observed in individuals with intellectual and developmental disability (Sloneem, 2005).

A number of studies have reported on the prevalence of specific forms of self-injury displayed by individuals with SMS. For example, Finucane et al (2001), using a measure developed specifically for the study, reported on the prevalence of the following behaviours; head-banging (55.2% of sample), hand/wrist biting (93.1%), skin picking (51.7%), slapping self (62.1%), hairpulling (34.5%) polyembolokomania (inserting objects in ears 31%; nose 17.2%; rectum 3.5%; vagina 21.1%) and onychotillomania (pulling out fingernails 48.3%; pulling out toenails 34.5%). Dykens et al (1998), using a modified version of the SIB-C, reported that self-biting and self-hitting were the most frequent reported topographies of self-injury, occurring in 77% and 71% of their sample respectively. Less common behaviours included nail yanking (29% of sample) and inserting foreign objects into body (25%). A more recent study reported by Martin et al (2006), again using the modified SIB-C, noted the following behaviours in children with SMS; hitting self (93% of sample), self-biting (80%), hitting self against objects or surfaces (53%), inserting fingers or objects into body openings (47%), pulling hair or skin (40%), rubbing/scratching self (33%), pulling hair out (27%), hitting self with object (27%), eye-poking (20%), pulling out finger or toe nails (13%), pulling out teeth (7%).

Stereotypical behaviour.

SMS appears to be associated with a range of stereotypical behaviours, some of which are apparently unique to the syndrome. In their recent meta-analysis, Edelman et al (2007) reported that 89.7% of individuals with SMS were reported to have displayed stereotypical behaviour. Finucane, Konar, Haasgivler, Kurtz and Scott (1994) reported 11 individuals with SMS (aged 7-51 years) who presented with a highly characteristic 'spasmodic upper body squeeze'. Dykens and Smith (1998) using a modified version of the *Stereotypy Checklist* (Bodfish et al., 1995) reported that their entire sample displayed at least one topography of stereotypical behaviour. The most frequent of which were inserting hands (69% of sample) and objects (54%) into mouth and teeth grinding (54%). Self-hugging and 'lick and flip' repetitive page turning were recorded in 46% and 51% of the sample respectively. Martin et al (2006), using the same measure, reported on the prevalence of various topographies of stereotypical behaviours. Commonly occurring topographies included: inserting hand into mouth (87% of sample), inserting object into mouth (80%), covering ears/eyes (67%), hugs/squeezes upper body (60%), 'lick and flip' repeatedly turning of book pages (60%), hand-flapping/waving (60%), purposefully dropping/throwing objects (60%).

Aggression and property destruction.

There is some evidence to suggest that individuals with SMS may be especially likely to display aggressive behaviours. In their between-group comparison, Dykens and Smith (1998) report that children with SMS scored significantly higher on the aggressive behaviour sub-domain of the *CBCL*, than did children with Prader-Willi syndrome or a mixed intellectual disability control group. Some 57% of their sample was reported to display physical aggression and 86% displayed property destruction. Dykens et al (1997) report that 7 of the 10 individuals in their sample (aged 14-51 years) displayed aggressive behaviours as measured by the *Reiss Screen* (S. Reiss, 1988). Sloneem (2005) reported that 81.3% of individuals with SMS in her sample displayed property destruction and 87.5% physical aggression. Individuals with SMS displayed significantly more pinching, biting, hitting, grabbing, kicking, head-butting, hair-pulling, choking and using objects as weapons than that reported in the literature for people with intellectual and developmental disability generally.

Summary

Behavioural phenotype research has represented a search for associations between genetic syndromes and various topographies of behaviours. Both FXS and SMS appear to be associated with a characteristic pattern of challenging behaviour. The extent to which such behaviours can be considered genetically determined remains somewhat contentious. Whilst such behaviours are often considered 'phenotypic' of these syndromes, it should be noted that environmental factors, such as the quality of home environments and the effectiveness of therapeutic interventions, have been shown to influence the occurrence of such behaviours (Hall et al., 2009; Hessl et al., 2001). In addition, secondary genes may also be an important contribution to behaviours displayed by individuals with single gene disorders, such as FXS and SMS. For example Hessl et al (2008) reported that a functional polymorphism in the serotonin transporter 5-HTTLPR influenced the extent to which males with FXS were reported to display stereotypical, aggressive and destructive behaviours. In light of this evidence the proposition that a single genetic abnormality determines challenging behaviour in either syndrome appears to be difficult to uphold.

Chapter Summary and Thesis Overview

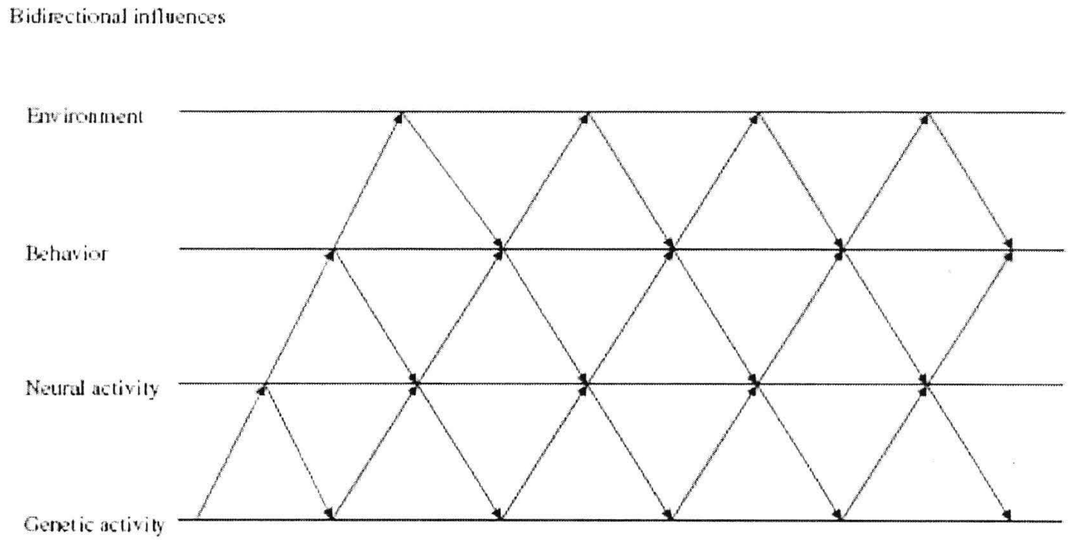
Challenging behaviour has been introduced as a socially significant problem. Given the pervasive negative impact of challenging behaviour on the quality of life of those who display it, as well as its prevalence and relative persistence in the absence of effective intervention, it would seem to merit continued investigation. Applied behaviour analysis has led to the development of methods specifically designed to identify the environmental factors that influence the occurrence of challenging behaviour. Challenging behaviour has been consistently shown to belong to at least one of four functional classes. In contrast, the behavioural phenotype approach has emphasised the importance of genetic factors in the causation of challenging behaviour. Studies have reported associations of varying specificity between genetic syndromes, such as FXS and SMS, and certain topographies of challenging behaviour.

The analysis of the role of genes and environment in challenging behaviour has, with only a few exceptions, taken place in relative isolation of the other. There has been scant attention paid to potential interactions between genetic and environmental sources of variability. The aim of this thesis is to provide both a conceptual and empirical examination of the interaction between such variables in the development and maintenance of challenging behaviour.

The overarching position of the current thesis is that as with all behaviour, challenging behaviour forms part of a developmental system (Gottlieb, 1992, 2003; Johnston & Edwards, 2002). That is, challenging behaviour occurs within a biological and environmental context (Morris & Midgley, 1990; Thompson, 2007). Such a system, as depicted in Figure 1.1, is comprised by multiple bi-directional relations between variables at different levels of analysis, in which each influences and is influenced by the other. The exclusion of certain variables, whether they are genetic or environmental, fails to provide a complete account of behavioural development as it occurs within the developmental system. The primary focus of the current thesis is on how such bi-directional influences serve to establish the motivation for those consequences that maintain challenging behaviour.

Chapters 2-5 provide both a conceptual and empirical examination of this hypothesis. It is proposed that gene-environment interactions (GxE) may hold important implications for the understanding of challenging behaviour in people with intellectual and developmental disabilities. Evidence is provided to suggest that one role of GxE may be to alter the value of certain socially- and non-socially mediated events as types of reinforcement or punishment.

Figure 1.1. A developmental systems model (from Gottlieb, 1992, p. 186).



Chapter 2 provides a systematic review of the concept of the 'motivating operation' (Laraway, Snycerski, Michael, & Poling, 2003). The review provides a critical analysis of the literature on motivating operations as applied to the functional analysis of challenging behaviour from 1998-2007. The review functioned as a scoping exercise with which to identify potential areas for future research and to inform subsequent conceptual and empirical work. One area that is suggested as requiring further research lies in the interaction between genetic and environmental variables.

Chapter 3 narrows the focus to the analysis of GxE and specifically examines how such relations influence the early development of self-injurious behaviour, as well as other challenging behaviours. The chapter aims to provide an account of why certain genetic syndromes are associated with particular 'phenotypic' behaviours. This analysis extends existing accounts of the genesis of challenging behaviours and argues that further research is required to investigate the nature of such interactions.

Chapters 2 and 3 both postulate that genetic factors may influence challenging behaviour by altering the reinforcing or punishing value of certain stimuli. The empirical part of this thesis aimed to provide an examination of this hypothesis by determining whether there were both within- and between-syndrome differences in the function served by challenging behaviour.

Chapter 4 provides an indirect examination of this question. Parents of children with FXS and SMS were interviewed, as was a 'mixed etiology' control group using an indirect measure of behavioural function. The groups were shown to differ in the probability of children being reported as displaying challenging behaviours that served particular functions.

In chapter 5, experimental analogue methodology was used to address this same question. Using a subset of participants from the previous study, this involved comparing the results of functional analyses conducted with a group of children with FXS against those conducted with a group of children with SMS. The groups appeared to differ in a manner consistent with that found in the indirect study.

Finally, in chapter 6 the findings of the current thesis are related to the underlying literature and the implications for future research and the understanding, assessment and treatment of challenging behaviour are discussed.

Chapter II. Motivating Operations and Challenging Behaviour. A Systematic Review[7]

"It seems likely that clinically significant, long-term, generalized change in problem behaviour can occur only if the EOs that evoke problem behaviour are, directly or indirectly, addressed in treatment." (McGill, 1999, pp. 406-407)

[7] Adapted portions of this chapter have been submitted to the *Journal of Applied Behavior Analysis* and *Behavior Analysis in Practice* for review.

Chapter Overview

Motivating operations (MOs) refer to antecedent events that alter the *value* of those consequences that serve to maintain or punish operant behaviour and alters the probability of *behaviour* occurring that has been previously associated with those consequences. Such variables have notable implications for the understanding of challenging behaviour. The current chapter provides a systematic review of the literature on MOs and the functional analysis of challenging behaviour from 1998-2007. The literature related to MOs for social positively reinforced, social negatively reinforced and automatically reinforced challenging behaviours is reviewed. The chapter provides a synthesis of developments in the literature on MOs and functional analysis since 1998. A number of areas in which future research on MOs is required are identified. This process functioned as a scoping exercise to guide the remainder of the thesis. Of particular relevance is the suggestion that further work be conducted that delineates the motivative influence of genetic events on challenging behaviour.

Introduction

The original emphasis of functional analysis lay on the role of behaviour-consequence relations. Over the past fifteen years there has been an increased appreciation of the role that antecedent events can have on challenging behaviour. Indeed, antecedent variables, such as motivating operations (MOs), must be incorporated into functional analysis in order to fully understand the influence of behaviour-consequence relations. MOs refer to antecedent events that alter the value of those consequences that maintain or punish operant behaviour, and alter the probability of associated behaviours occurring. The current chapter provides a systematic review of the literature on MOs as applied to the assessment of challenging behaviour from 1998-2007. The aim of this process is to act as a scoping exercise to identify avenues for future research. Areas that merit further investigation are proposed. Most importantly for the current thesis is the suggestion that further work is required to explore interactions between behaviour-environment relations and various biological influences. A number of suggestions along these lines are proposed.

Almost a decade ago McGill (1999) examined the implications that the concept of the establishing operation (EO) held for the assessment, treatment and prevention of challenging behaviour. This paper, together with the more general review of antecedent events by Smith and Iwata (1997), reflected a shift in emphasis for the applied analysis of challenging behaviour. This shift has led to a greater appreciation of the role played by motivative variables in challenging behaviour (J. Michael, 2000). In the years since the publication of these papers, there have been considerable advances in our understanding of such variables. These advances have been multifaceted, occurring at both the conceptual and empirical level. For example, the term MO has now subsumed that of the EO. The concept of the MO has been successfully applied to areas as diverse as organizational behaviour management (Olson, Laraway, & Austin, 2001), eating disorders (Tapper, 2005), and basic research on bio-behavioural relations (Kennedy, 2002).

These advances are perhaps best characterised by the increase in the number of papers to include the discussion of MOs within the pages of the *Journal of Applied Behaviour Analysis* (JABA). Figure 2.1 shows the number of papers within *JABA* to have cited one of the four main papers on MOs by Michael and colleagues (Laraway, Snycerski, Michael, & Poling, 2003; Michael, 1982, 1993, 2000), as well as all papers within *JABA* published since 1982 which have included the terms "establishing operation", "abolishing operation" or "motivating operation"[8]. Figure 2.1 shows that there has been continued exponential growth in the number of papers that relate to MOs. Over 60% of these papers have been published in the last 10 years.

Given the substantial developments that have taken place since the publication of the Smith and Iwata (1997) and McGill (1999) reviews it seems that a quantitative and qualitative synthesis of this work as applied to the functional analysis of challenging behaviour is warranted. This process will serve three purposes. First, it will prompt some reflection on how far the science has come in incorporating once neglected variables into the functional analysis of challenging behaviour. Second, in accordance with one of the founding principles of applied behaviour analysis (Baer, Wolf, & Risley, 1968), it will aim to provide a conceptual synthesis of a broad range of research on the MO, relating diverse empirical findings to potential underlying behavioural principles. Finally, it will serve to identify areas where future research can begin to shed new light on our understanding of MOs and challenging behaviour and thereby identify potential areas of research for the current thesis.

[8] Papers that used terms other than these i.e., "disestablishing operations", "establishing conditions" were excluded from this analysis, as were papers for whom the terms were only found in the reference list.

Motivating Operations

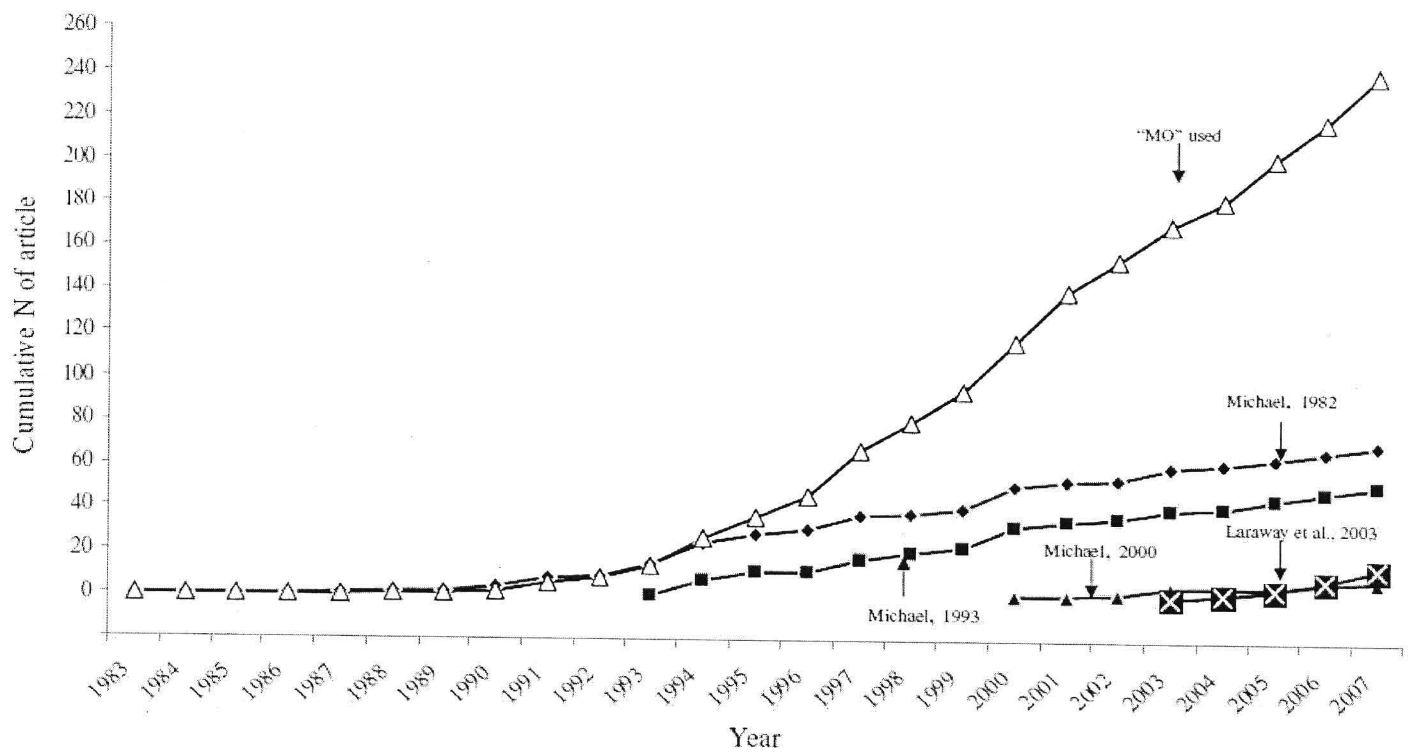
Within the operant model it has been long established that the value of a given consequence as a type of reinforcement or punishment is in constant flux, as is the probability of behaviour occurring that has previously been associated with such consequences (Fuller, 1949; Skinner, 1953). A systematic approach to the analysis of such events was absent however leaving "a gap in our understanding of operant functional relations" (Michael, 1993, p. 191). This gap was addressed by Jack Michael (1982; , 1993) who cogently argued for a distinction to be made between antecedent stimuli that were correlated with the availability of reinforcement or punishment (discriminative stimuli) and antecedent stimuli that altered the value of such consequent events (EOs).

Laraway et al. (2003) recently refined the concept, proposing that the omnibus term of the *motivating operation* replace that of the EO. The term MO is the product of an evolution in operant terminology. Michael (1982) originally used the term *establishing operation* to refer to antecedent events that momentarily alter a) the effectiveness of consequent events that function as reinforcers or punishers (termed the *reinforcer-establishing* effect) and b) the frequency of responses that have been associated with those consequences in the past (termed the *evocative-effect*). For convenience, movements in the opposite direction were subsumed within the rubric term of the EO. As Michael (1982) states "'establishing' should be taken to be short for 'establishing or abolishing'." (p.151). The use of the term EO in this manner, however, did not adequately capture the bidirectional effects of motivating events potentially restricting our ability to predict and control behaviour (Laraway, Snycerski, Michael, & Poling, 2003).

MOs refer to antecedent events that share two main properties. The first property termed the *value-altering effect*, refers to the effect of an antecedent event on the effectiveness of other stimuli that function as types of reinforcement or punishment (Laraway, Snycerski, Michael, & Poling, 2003). An EO establishes the effectiveness of a particular type of reinforcement or punishment, whereas an AO abolishes the effectiveness of a particular form of reinforcement or punishment. For example, for an athlete the value of a drink is established as an effective reinforcer following a long run; whereas it is abolished after he has consumed a large quantity of water.

The second property of the MO is referred to as the *behaviour-altering effect* (Laraway, Snycerski, Michael, & Poling, 2003). Thus an EO evokes behaviour that has been previously associated with the events it establishes as reinforcers (and vice-versa for behaviours associated with punishment), whereas an AO abates behaviour that has been associated with events it abolishes as reinforcers (and vice-versa for behaviours associated with punishment). Taking the same example, an athlete following a long run will be more likely to display behaviours that have in the past been associated with a drink, such as walking to the drinks machine. Likewise, after drinking a large quantity of the same drink the probability of the athlete displaying the same behaviour abates. Michael (1982) notes that the MO may alter behaviour either directly, or indirectly for example, by altering the evocative effect of discriminative stimuli or by influencing the value of conditioned reinforcers or punishers. Descriptions of the MO have typically been restricted to changes in frequency however, as Michael (2007) notes, the MO may alter other dimensions of behaviour such as response latency, magnitude or relative frequency.

Figure 2.1. Cumulative number of articles published in *JABA* in which the term MO, EO or AO have been used or in which citation to Michael's (1982, 1993, 2000) articles or Laraway et al's (2003) article has appeared.



Laraway et al (2003) did not include a discussion of the subtypes of the MO in their review. As such a description of each subtype of MO will be provided in light of terminological changes that have occurred since the recommendations made by Laraway et al (see also Michael, 2007).

Unconditioned MOs

Certain events may acquire their value- and behaviour-altering effects as a result of the organism's phylogenic history and such events are termed unconditioned MOs (UMOs). Deprivation of primary types of reinforcement, such as food, drink, or sexual activity, establishes the value of these stimuli as reinforcers and exerts an evocative effect on associated behaviours, just as satiation of these types of stimuli abolishes their reinforcing value and abates associated behaviours. Deprivation and satiation also influence the extent to which the contingent removal of primary types of reinforcement will function as a type of punishment. Thus, a person must be deprived of food (or in common parlance be 'hungry') in order for the contingent removal of food to act as an effective type of punishment. The onset of certain forms of aversive stimulation, such as pain or temperature changes above or below a comfortable level, similarly acts as a UMO for negatively reinforced behaviour. Whilst the behaviours that these events alter the frequency of are often learnt the source of 'motivation' for the consequences that they maintain is not.

Conditioned MOs

Other events acquire their value-altering effects as a result of the organism's ontogenic history. Such events are termed conditioned motivating operations (CMOs). Previously neutral events may acquire the status of a CMO after having systematically preceded or having been paired with a UMO, another CMO, or particular types of reinforcement or punishment. Three forms of CMO have been proposed; *surrogate CMOs*, *reflexive CMOs* and *transitive CMOs*.

A surrogate CMO (CMO-S) is a previously neutral stimulus that following temporal association with a UMO or other CMO independently alters the effectiveness of other stimuli as reinforcers or punishers and alters the probability of associated behaviours. Take for example, a person who always has their lunch at midday, the time on the clock in addition to having discriminative properties (such as signalling the opening of the canteen) may also exert a motivative influence. Following the repeated pairing of food deprivation (the UEO) and the time of 12pm on a clock, the time on the clock may itself establish the reinforcing value of food and evoke food-related behaviour independent of actual levels of food deprivation. The time on the clock may also establish the punishing value of food unavailability and abate behaviours that have been associated with such delays in the past, such as answering the telephone. Similar effects could occur in the opposite direction. Take for example, stimuli that are temporally associated with being awake (such as it being light outside, birds singing, the smell of coffee being brewed downstairs etc). The onset of such stimuli may abolish the reinforcing value of continued sleep and abate sleep-related behaviours independent of actual levels of sleep deprivation. Sleep is no less or more available in such a situation, but is less reinforcing.

Michael described the reflexive CMO (CMO-R) as constituting a 'promise' or 'threat' CMO. That is, the onset of a CMO-R is associated with either the *improvement* or *worsening* of the organism's condition. Therefore, its onset alters the value of its own removal (or continued presence) as a type of reinforcement (or punishment) and alters the probability of behaviours occurring that have previously been associated with these consequences. The CMO-R therefore acts on its own reinforcing value and not on that of another stimulus (as in the surrogate CMO). Take an amateur runner for whom the onset of certain stimuli (such as a large-looking hill in the near distance) is correlated with the onset of physical discomfort, such as a stitch or cramp, and thus the 'worsening' of his condition. The sight of the hill may establish its own offset as an effective form of reinforcement and evoke behaviours that have been associated with its removal in the past, such as taking a different route that avoids the hill (acting therefore as a CEO-R). In contrast, for a more experienced runner, (for whom physical discomfort functions as a conditioned reinforcer) the onset of the hill may be correlated with the release of endorphins and therefore the 'improvement' of his condition. The onset of the hill therefore establishes its continued presence as an effective type of reinforcement and evokes behaviours that have led to this in the past, such as continuing to run in the direction of the hill. Similar effects may apply with punishment. For example, for the more experienced runner the onset of the hill would be likely to establish its offset as an effective type of punishment. When would an event or stimulus change constitute a CAO-R? Take the amateur runner for whom the onset of the hill acts as a CEO-R and evokes behaviour that is successful in avoiding the hill. As the presence of the hill acts on its own reinforcing value, it is the absence of the hill (for example, the disappearing sight of the hill or the sight of flat ground) that functions as a CAO-R.

A transitive CMO (CMO-T) refers to stimuli in whose context the value of existing conditioned reinforcers or punishers is altered, as is the likelihood of behaviours occurring that have been associated with such consequences in the past. Think of an individual with intellectual and developmental disabilities who lives in a residential home where access to food in the fridge is restricted by a lock on the fridge door. In such a situation, the response of independently opening the fridge door and subsequent reinforcement, in the form of access to food, is unavailable. This may establish the presence of someone who can open the fridge door (i.e., a member of staff with the key) as an effective source of reinforcement and evoke behaviours that have led to this in the past (i.e., manding or aggression). The member of staff and the key are no more available in the presence of the locked fridge but are more reinforcing. In the absence of the lock on the fridge the reinforcing value of the member of staff with a key is abolished and behaviours, such as manding or aggression, abate. Similar effects may occur for conditioned types of punishment. For example, think of a school child for whom time-out is imposed contingent on disruptive behaviours during class. The value of time out as a type of punishment is more likely to be established in the presence of certain stimuli, such as the lesson involving a highly preferred activity, than in the presence of others, such as the lesson involving a non-preferred activity.

One benefit to have emerged from the recent developments in terminology surrounding motivative events has been the explicit recognition that MOs may have multiple and simultaneous effects (Laraway, Snycerski, Michael, & Poling, 2003). That is, the abolishing and abative effects of the AO are of equal importance as are the establishing and evocative effects of the EO. As implied in some of the examples above a single event may function as both an EO and an AO for different behavioural consequences. For example, providing high levels of noncontingent attention may abolish the reinforcing value of attention, but may also establish the reinforcing value of toys used for solitary play. Likewise a single behaviour may be influenced by more than one MO. For example, the *abolishing* and *abative* effects of non-contingent escape may also be influenced by other MOs such as the amount of sleep the individual has had, the difficulty of the request and so on. Such complexity is perhaps better encapsulated by the term MO. Michael (2007) also notes that a single stimulus change may exert not only value- and behaviour-altering effects but also function-altering effects (i.e., by operating as a type of reinforcement or punishment). The deprivation of attention, for example, will function as an EO for subsequent attention-maintained behaviour but will also function as a type of punishment for the behaviour that precedes the onset of deprivation. Likewise, providing access to a food tangible will reinforce food-maintained behaviours but may also serve to abolish the effectiveness of the food tangibles and abate subsequent behaviour maintained by access to food. It is important that such relations are applied to the study of challenging behaviour and it would seem that the term MO better encapsulates such complex relations than that of the EO.

Method

Studies relevant to MOs were identified using a three-stage process. First, papers that were published between 1998-2007 which cited at least one of the four main papers by Michael and colleagues (Laraway, Snycerski, Michael, & Poling, 2003; Michael, 1982, 1993, 2000) were identified via the database *Web of Science*. The four journals that had the highest number of citations in this initial search (*Journal of Applied Behaviour Analysis*, *Research in Developmental Disabilities*, *Behavioural Interventions* and *American Journal on Mental Retardation*) were then hand-searched for the years 1998-2007. The reference section of each article was then examined to identify any additional MO studies. Finally all studies were reviewed to see if they met criteria for inclusion in the present review.

Inclusion and Exclusion Criteria

All studies were empirical and involved the assessment of MOs as applied to challenging behaviour. All studies involved some form of functional assessment, including indirect assessments (Cuvo, May, & Post, 2001), AB functional analyses (e.g., Carr & Durand, 1985) and ABC functional analyses (e.g., Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994). Studies on automatically reinforced challenging behaviour were included that examined challenging behaviour which persisted in the absence of social consequences (e.g., Goh, Iwata, & Kahng, 1999). All studies relied on the direct observation of target behaviours. All studies involved experimental manipulation and purely descriptive studies were excluded from analysis. Studies predominantly included participants with intellectual and developmental disabilities;

however all studies that met the inclusion criteria and had participants of typical intellectual functioning were also included. Studies that solely involved an evaluation of treatments that have been previously analysed in terms of the MO (such as behavioural momentum or instructional fading) and that did not have additional implications for the assessment of challenging behaviour were generally excluded from review. Where there were clear implications for the role of MOs in the assessment of challenging behaviour then the study was included. Papers were excluded if they did not include the assessment or manipulation of MOs or were not focused on challenging behaviour. The papers allow, therefore, for a systematic review of research over the last 10 years on the role of MOs in the functional analysis of challenging behaviour.

Results

Using this method a total of 95 empirical studies were identified that met the inclusion criteria.

Table 2.1 shows the journals to have published empirical studies that met the inclusion criteria. A total of 11 journals published at least one study that met the inclusion criteria. Only five journals have published more than two papers that met the inclusion criteria. The largest number of studies involving MO manipulations was published in the *Journal of Applied Behaviour Analysis*. The four journals that were hand searched (*Journal of Applied Behaviour Analysis*, *Research in Developmental Disabilities*, *Behavioural Interventions* and *American Journal on Mental Retardation*) provided some 91% of the total sample of studies included in the review.

Table 2.1.

Number and Percentage of MO Studies that Met Inclusion Criteria by Journal Name

Journal name	No. of studies	% of sample
Journal of Applied Behavior Analysis	55	57.9%
Behavioral Interventions	17	17.8%
Research in Developmental Disabilities	11	11.5%
American Journal on Mental Retardation	3	3.1%
Behavior Modification	3	3.1%
Journal of Positive Behavioral Interventions	1	1%
Education and Treatment of Children	1	1%
Journal of Developmental and Physical Disabilities	1	1%
Journal of Early and Intensive Behavior Intervention	1	1%
School Psychology Quarterly	1	1%
School Psychology Review	1	1%

Motivating Operations and Functional Analysis

Functional analysis provides a powerful means of identifying the consequences that maintain challenging behaviour (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994). The variables that maintain challenging behaviour are typically categorised into four general classes of reinforcement; *social-positive reinforcement, social-negative reinforcement, positive automatic reinforcement* and *negative automatic reinforcement*. For each consequence that maintains challenging behaviour there is a parallel MO which alters the value of that consequence as a type of reinforcement (McGill, 1999). The experimental literature to have examined the influence of MOs on the occurrence of challenging behaviour maintained by each reinforcement contingency is now systematically reviewed. For each general class of reinforcement[9] an analysis of how the research literature has evolved over the past decade is provided. Important areas for future research are subsequently discussed.

Social-Positive Reinforcement

Attention.

Challenging behaviour, such as self-injury or aggression, may be maintained by the contingent provision of social attention (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994). Of the 536 functional analyses included in a recent review of the literature, 25.3% were interpreted as showing a pattern of responding consistent with such a hypothesis (Hanley, Iwata, & McCord, 2003). Whilst such behaviours are maintained by attention, the value of attention varies across different environmental contexts. It seems likely that the environments in which many individuals with intellectual and developmental disabilities reside are replete with characteristics that serve to establish attention as an effective reinforcer and as such an analysis of MOs is likely to shed important light on current understanding of attention-maintained challenging behaviour.

A decade ago the evidence base for the influence of MOs on attention-maintained challenging behaviour was relatively sparse. In his review McGill (1999) suggested that challenging behaviour maintained by access to attention was particularly likely to occur in situations characterised by low levels of social contact. A series of studies that supported this interpretation were cited. These studies relied on either within-session antecedent manipulations, such as that found in the standard attention-condition of a functional analysis (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994), or descriptive analysis (Hall & Oliver, 1992; Taylor & Carr, 1992). Several

[9] Due to the lack of research on behaviours that serve a non-socially mediated (automatic) negatively reinforced function, the following considers only the following classes of reinforcement: *social-positive reinforcement*, *social-negative reinforcement*, and *automatic reinforcement*.

studies were also cited which reported the abolishing and abative effects of providing dense levels of social contact on a fixed time schedule (e.g., Hagopian, Fisher, & Legacy, 1994; Vollmer, Iwata, Zarcone, Smith, & Mazaleski, 1993). It was also recognised that other variables, such as providing non-contingent access to arbitrary reinforcers, such as toys, altered the probability of attention-maintained challenging behaviour (e.g., Fischer, Iwata, & Mazaleski, 1997).

Thus, it was well established that momentary changes in the level of social contact could influence attention-maintained challenging behaviour. The majority of these studies relied on either descriptive or within-session experimental manipulations. Whilst it was recognised that other variables could exert value- and behaviour-altering effects on attention-maintained challenging behaviour, their examination remained at a preliminary stage.

Great strides have been made in the past decade regarding our understanding of the variables that evoke attention-maintained challenging behaviour. The current review identified some 21 studies published since 1998, which involved the assessment of MOs for attention-maintained challenging behaviour (see Appendix 1).

Investigators have continued to demonstrate that within-session variation in levels of social contact may act on the value of attention as a type of reinforcement (Roane, Lerman, Kelley, & Van Camp, 1999; Worsdell, Iwata, Conners, Kahng, & Thompson, 2000). Worsdell et al (2000) found that the attention- or tangible-maintained self-injury of six individuals occurred at consistently high rates only in those conditions in which both an EO (e.g., within-session deprivation of attention) and reinforcement contingency were present. Such findings suggest that momentary changes in the levels of attention during a functional analysis exert value- and behaviour-altering effects, and that this is particularly likely to be observed when a reinforcement contingency is also present.

A total of eight studies were identified that examined the influence of systematic manipulation of pre-session levels of social contact for attention-maintained challenging behaviour (Berg et al., 2000; McComas, Thompson, & Johnson, 2003; O'Reilly, 1999; O'Reilly, Edrisinha, Sigafoos, Lancioni, & Andrews, 2006; O'Reilly, Edrisinha, Sigafoos, Lancioni, Machalicek et al., 2007; O'Reilly, Lancioni, & Emerson, 1999; O'Reilly et al., 2006; Roantree & Kennedy, 2006). McComas et al (2003), for example, demonstrated that providing continuous pre-session social contact reduced subsequent challenging behaviour in the attention condition of a functional analysis, whereas challenging behaviour increased following pre-session conditions in which social contact was unavailable. Interestingly, manipulating pre-session levels of social contact had no influence on the escape-maintained challenging behaviour of two children. These methodological developments, in which prior access to putative maintaining variables are systematically manipulated, have allowed for a greater degree of experimental control over 'third variables' than has previously been the case and have provided a foundation on which more complex relations can begin to be investigated.

These studies hold important implications for the assessment of attention-maintained challenging behaviour and specifically in relation to the sequencing of experimental conditions during a functional analysis. It seems likely that attention-maintained challenging behaviour will occur at higher rates following an alone condition and at substantially lower rates following a play or demand condition which are typically characterised by high rates of attention, providing some empirical support for the Iwata et al (1994) recommendation that functional analyses be sequenced in a semi-random manner. Such findings also imply that pre-existing environmental conditions can exert a powerful influence over behaviour in a subsequent functional analysis. Thus, it is possible that responding within a functional analysis tells us as much about pre-existing environmental factors as about the influence of behaviour-environment contingencies within the functional analysis. This highlights the importance of establishing control over pre-session events that may influence subsequent behaviour-environment relations in a functional analysis.

The MO has often been treated as a dichotomous variable, whereby an EO is judged as being either present or absent based on a given experimental manipulation (e.g., Worsdell, Iwata, Conners, Kahng, & Thompson, 2000). In the case of attention-maintained challenging behaviour the absence of social contact is typically referred to as an EO and access to social contact as an AO. A recent study by Roantree and Kennedy (2006), however, highlights the importance of providing a functional, as opposed to a procedural definition of the MO. Providing access to pre-session attention revealed a clear attention function for the stereotypical behaviour of a boy after stereotypy appeared undifferentiated in an initial analysis. The authors suggest that providing access to attention in pre-session conditions served as an EO rather than as has typically been demonstrated, an AO. Such an effect may parallel 'reinforcer sampling' procedures which are used to establish 'motivation' for tangible items (Ayllon & Azrin, 1968; Mueller, Wilczynski, Moore, Fusilier, & Trahant, 2001). Such findings, whilst in need of experimental replication, indicate that the MO rather than being a dichotomous variable, as is sometimes implied, may be better conceptualised on a continuous scale, the definitions of which should be based on observed changes in behaviour as opposed to procedural manipulations.

The manipulation of variables other than social contact may also act as an MO for attention-maintained challenging behaviour. For example, the availability of preferred tangibles in the attention condition of a functional analysis may act as an AO for attention-maintained challenging behaviour (e.g., Fisher, O'Connor, Kurtz, DeLeon, & Gotjen, 2000; Ringdahl, Winborn, Andelman, & Kitsukawa, 2002)[10]. The relative

[10] See McCord and Neef (2005) for a discussion of the implications of such effects for functional analyses.

preference for tangibles appears to be a particularly important variable, with high preferred items more likely to reduce the occurrence of attention-maintained challenging behaviour than are less preferred items (Fisher, O'Connor, Kurtz, DeLeon, & Gotjen, 2000). It has been proposed that the presence of highly preferred items abolishes the reinforcing value of attention and abates attention maintained challenging behaviour. Given that the attention condition of a standard functional analysis involves providing access to toys/activities it would seem an important step to ensure that such toys are not so highly preferred that they reduce the value of attention below a level that would be typically expected in the absence of the toy/activity. One could also envisage a situation whereby the presence of certain toys functions as an EO for attention-maintained challenging behaviour. For example, the presence of a toy that cannot be independently operated may heighten the reinforcing value of attention and evoke challenging behaviour, by acting as a transitive CEO. However, no studies were identified in the current review that demonstrated such relations for attention-maintained challenging behaviour.

Approximately 4.1% of functional analyses result in an undifferentiated pattern of responding (Hanley, Iwata, & McCord, 2003), making a judgement about behavioural function difficult to ascertain. In some cases such behaviours may be socially mediated but the functional analysis fails to capture this, resulting in a false negative result (Carr, Yarbrough, & Langdon, 1997). Changes made to *whom*, *where* and *how* a functional analysis is conducted may all act as MOs and exert a significant influence over attention-maintained challenging behaviour.

The use of reversal designs with an embedded multi-element component has facilitated the investigation of such idiosyncratic variables. Indeed 40% of the studies presented in Appendix 1 utilised this form of experimental design. The advantages of such a design are relatively apparent, not only do they enable an analysis of the *main* *effects* of aspects of the current context (e.g., the influence of location on the overall occurrence of challenging behaviour) but also the *simple effects* of the functional analysis (e.g., attention-maintained behaviour across locations) and where present *interaction effects* between the two (e.g., a change in function across different locations).

Studies have shown that the occurrence of attention-maintained challenging behaviour may be influenced by the person who is running the functional analysis (English & Anderson, 2004; LeBlanc, Hagopian, Marhefka, & Wilke, 2001; Ringdahl & Sellers, 2000). Ringdahl and Sellers (2000) demonstrated that the attention-maintained behaviour of two individuals only became apparent when the functional analysis was run by the child's caregiver and was not apparent when sessions were run by clinic staff. Other variables such as the gender of the therapist have been shown to exert similar effects (LeBlanc, Hagopian, Marhefka, & Wilke, 2001). The presence/absence of specific individuals may act on the reinforcing value of attention and evoke or abate attention-maintained behaviour accordingly. For example, parental attention may hold a greater reinforcing value than that of other individuals. An alternative account could be that the presence of certain individuals (such as caregivers) is discriminative for the contingent provision of attention due to the history of reinforcement associated with that person[11]. It would be of interest for further research to begin to examine the mechanisms that underpin such relations.

The location in which a functional analysis is conducted can influence the occurrence of challenging behaviour and judgements that are made about the function served by that behaviour. For example, Harding, Wacker, Berg, Barretto, and Ringdahl (2005) reported that changing the location in which a functional analysis was conducted

[11] It could also be argued that these manipulations refer to different types of reinforcement (i.e., the attention of a parent is a different stimulus than the attention of another individual) and that these effects refer to changes in the type of reinforcement rather than the effects of MOs per se.

revealed a social function to challenging behaviour that had previously appeared undifferentiated across all conditions. Again it remains unclear what mechanism underpins such relations and whether location acted on the reinforcing value of social consequences or whether certain locations were simply discriminative for subsequent reinforcement. It seems important, however that this information be elucidated. A component analysis in which different aspects of a location are manipulated may enable further investigation of such variables. Identifying the specific aspects of a location that are associated with low rates of challenging behaviour would provide an obvious first step for subsequent intervention.

The EO in a standard attention condition of a functional analysis involves the within-session deprivation of attention, typically with the therapist withholding social contact by pretending to do some work. However, this arrangement is not always sufficient to precipitate challenging behaviour maintained by social attention and in some cases alternative antecedent manipulations are required (Call, Wacker, Ringdahl, & Boelter, 2005; O'Reilly, Lancioni, King, Lally, & Dhomhnaill, 2000; Ringdahl, Winborn, Andelman, & Kitsukawa, 2002). Call et al (2005) demonstrated that the attention-maintained challenging behaviour of one individual was evoked at consistently high levels only when a diverted attention condition was combined with a demand whilst challenging behaviour was reinforced by the contingent provision of attention. It may be that the presence of the demand established the reinforcing value of attention by functioning as a transitive CEO. As a parallel to Michael's (1982, 1993) slotted screw example, one could envisage that the onset of a demand that an individual cannot independently complete (= the sight of the slotted screw), establishes social contact (= the screwdriver) as an effective type of reinforcement and evokes challenging behaviour that has led to attention in the past (= asking his assistant for the screwdriver). Indeed McGill (1999, p. 402) suggested that if demands were to function as a transitive

CEO for attention-maintained challenging behaviour then one would expect higher levels of such behaviour in the presence of demands than in the standard attention condition, a pattern of responding which corresponds to that reported by Call et al.

Other antecedent conditions have been shown to influence attention-maintained challenging behaviour, including the presence of toys in the attention condition of a functional analysis (Ringdahl, Winborn, Andelman, & Kitsukawa, 2002) and the use of a diverted attention condition (O'Reilly, Lancioni, King, Lally, & Dhomhnaill, 2000). Diverted attention may function as a form of surrogate CEO. One could imagine that situations that involve a caregiver interacting with another person are associated with momentary reductions in the level of social contact received by the child. Over time this event would be expected to acquire establishing and evocative properties, independent of actual levels of social contact. If diverted attention were to function as a surrogate CEO for attention-maintained behaviour, one would expect there to be higher rates of challenging behaviour occurring in the diverted attention condition than in the standard attention condition even if actual levels of attention were the same. Providing non-contingent attention during the diverted attention condition would be expected to reduce the occurrence of such behaviours, presumably through extinction of the CMO-S. This corresponds to the pattern of responding found in the O'Reilly et al (2000) study.

The consequences provided for one response class during a functional analysis may also act as an MO for challenging behaviours that are members of alternative response classes. Piazza, Hanley, Fisher, Ruyter, and Gulotta (1998) investigated the multiply controlled challenging behaviour of two individuals. Whilst the onset of demands evoked negatively reinforced challenging behaviour, the break (and resulting deprivation of attention that it imposed) evoked attention-maintained behaviour. Thus, high rates of challenging behaviour during a demand condition may reflect not only an escape function but also an attention-function. The analysis of challenging behaviour across within-session variations in MOs appears to provide one means of clarifying the function of apparently undifferentiated challenging behaviour (Roane, Lerman, Kelley, & Van Camp, 1999). For example if challenging behaviour is at least in part maintained by access to attention, one would expect a higher proportion of challenging behaviours to occur at times when demands (and social contact) are absent relative to when present, a pattern of responding consistent with that reported by LeBlanc, Hagopian, Marhefka and Wilke (2001).

Several different types of attention, aside from social disapproval, have been shown to influence attention-maintained challenging behaviour (e.g., DeLeon, Arnold, Rodriguez-Catter, & Uy, 2003; Kodak, Northup, & Kelley, 2007; LeBlanc, Hagopian, Marhefka, & Wilke, 2001; Piazza et al., 1999; Richman & Hagopian, 1999). Richman and Hagopian (1999) found that reprimands failed to function as an effective type of reinforcement for the attention-maintained challenging behaviour of two participants, whilst exaggerated attention and physical contact did. Such manipulations act on the value of attention as a type of reinforcement and deprivation or satiation of these specific forms of attention presumably functions as an MO for attention-maintained challenging behaviour. This suggests that not all forms of attention are of equal reinforcing value and that the type of attention provided during a functional analysis should be based on careful assessment and not an arbitrary 'one-size fits all' decision.

Over the past decade various biological influences have begun to be incorporated into the functional analysis of challenging behaviour (Langthorne, McGill, & O'Reilly, 2007). Such factors form part of the context in which challenging behaviour occurs and may influence the value of attention and the likelihood of attention-maintained challenging behaviour. Two studies were identified that investigated the influence of the drug *methylphenidate* (MPH) on attention-maintained challenging behaviour (Dicesare, McAdam, Toner, & Varrell, 2005; Northup et al., 1999). Both studies revealed interactions between MPH and specific analogue conditions. Dicesare et al (2005) reported that the attention-maintained challenging behaviour of a young man with intellectual and developmental disabilities reduced notably in conditions in which MPH was administered in comparison to placebo conditions. Such findings suggest both that the results of a functional analysis may help to guide the prescription of certain psychoactive medications in the treatment of challenging behaviour and also suggest that functional analyses may provide a means of testing the effectiveness of certain pharmacological interventions (Matson & Wilkins, 2008).

Our understanding of the influence of MOs for attention-maintained challenging behaviour has progressed significantly over the past decade, as have methods for the investigation of such effects. Investigators have successfully gone beyond within-session manipulations to isolate the effects of pre-session manipulations on attention-maintained challenging behaviour. This has facilitated the investigation of the mechanisms by which the MO influences challenging behaviour, such as the potential 'priming' effect of brief pre-session access to attention (e.g., Roantree & Kennedy, 2006). The value- and behaviour-altering influence of variables other than social contact, such as the presence of toys, has also been well documented. A number of parameters surrounding the implementation of a functional analysis have been shown to act as MOs for attention-maintained challenging behaviour. For example, the person conducting the analysis, the location where it is conducted and how it is conducted have all been shown to influence attention-maintained challenging behaviour. Finally, certain biological variables have been shown to act as MOs for attention-maintained challenging behaviour.

Tangibles.

Whilst potentially confounded by the provision of attention, it appears that some 10% of cases of challenging behaviour are maintained by access to preferred items or activities (Hanley, Iwata, & McCord, 2003). The value of tangibles fluctuates across environmental conditions and is clearly influenced by MOs. Given that many individuals with intellectual and developmental disabilities live in settings where access to tangibles is controlled by others it seems likely that MOs hold important implications for our understanding of tangible-maintained challenging behaviour.

A decade ago, McGill (1999) argued that variations in the availability of tangible items or activities altered their value as a type of reinforcement and the probability of tangibly maintained challenging behaviour. A small number of studies were cited that had shown the deprivation of tangibles could influence both tangible-maintained adaptive (Vollmer & Iwata, 1991) and challenging behaviour (Wacker et al., 1996). The non-contingent provision of tangible items was also known to reduce tangible-maintained challenging behaviour, most probably by acting as an AO (Marcus & Vollmer, 1996). Thus, whilst the evidence base remained relatively small, it had been demonstrated that the deprivation and satiation of tangibles could influence the subsequent occurrence of tangible-maintained challenging behaviours.

In comparison to attention-maintained behaviour the development in the experimental literature for tangible-maintained challenging behaviour has been relatively lean. The current review identified 16 studies that involved the investigation of MOs for challenging behaviour that was at least in part maintained by access to tangibles (see Appendix 2).

Momentary changes in within-session access to tangibles have been investigated in two studies as an MO for tangible-maintained challenging behaviour (Roane, Lerman, Kelley, & Van Camp, 1999; Worsdell, Iwata, Conners, Kahng, & Thompson, 2000), with both studies revealing higher levels of challenging behaviour following the within-session deprivation of the putative tangible maintaining variable, suggesting that this functioned as an EO for subsequent tangible-maintained behaviour.

Only two studies were identified that adopted pre-session manipulations to investigate tangible-maintained challenging behaviour (O'Reilly, Edrisinha, Sigafoos, Lancioni, Cannella et al., 2007; O'Reilly et al., 2006). As part of a wider study, O'Reilly, Sigafoos et al (2006) reported that the tangible maintained challenging behaviour of one young man occurred at higher rates following pre-session conditions in which there was no access to tangibles than in pre-session conditions in which the individual had free access to tangible items. Other studies have shown that such manipulations can exert powerful effects on an individual's preference for tangible items (e.g., Gottschalk, Libby, & Graff, 2000; McAdam et al., 2005) and the occurrence of tangible-maintained adaptive behaviours (e.g., Zhou, Iwata, & Shore, 2002).

Such findings hold important implications for the implementation of the tangible condition in a functional analysis. One would expect higher rates of tangible-maintained challenging behaviour following an alone or demand condition (in which access to tangibles are typically restricted) and lower rates following an attention or play condition (in which the individual typically has had non-contingent access to at least moderately preferred toys/activities). It is also likely that the amount of access an individual has had in their natural environment to preferred tangibles may influence the occurrence of tangible-maintained challenging behaviour within a functional analysis. As with attention-maintained challenging behaviour, this would suggest that prior access to tangibles should be controlled prior to conducting a functional analysis. Further studies are required that utilise pre-session manipulations in order to investigate the influence of the MO on tangible-maintained challenging behaviour.

Variables other than pre-session access to tangibles have been shown to influence tangible-maintained challenging behaviour, however. In their study, Ray and Watson (2001) investigated correlations between challenging behaviour and a series of 'temporally distant events'. The initially undifferentiated functional analyses of three school-age boys revealed a tangible function when analysed according to whether certain antecedent events, such as having less than 5 hours sleep, being woken late and nocturnal enuresis had occurred earlier in the day or not[12]. Such correlational evidence, whilst falling short of providing a 'believable demonstration' (Baer, Wolf, & Risley, 1968), may be important in generating hypotheses to account for otherwise unexplained variability in challenging behaviour during a functional analysis. For example, descriptive assessments may help in the identification of idiosyncratic variables that would not necessarily be detected using more formal experimental methods (Mace, Lalli, & Lalli, 1991).

As with attention-maintained challenging behaviour, it has been shown that relatively idiosyncratic contextual variables, such as the nature of the person running the functional analysis (McAdam, DiCesare, Murphy, & Marshall, 2004), as well as the location in which the analysis is conducted (Harding, Wacker, Berg, Barretto, & Ringdahl, 2005) can influence the occurrence of tangible-maintained challenging behaviour. Whether such effects are a function of the discriminative or motivative properties of such stimuli remains unclear, however, and further studies are required to elucidate such relations.

One critically important variable that has been shown to influence tangible-maintained challenging behaviour is the individual's preference for the tangibles withheld during a functional analysis (McLaughlin et al., 2003; Mueller, Wilczynski, Moore, Fusilier, & Trahant, 2001). In their study, Mueller et al (2001) reported tangible-maintained challenging behaviour as being more likely to occur in conditions in which access to higher-preferred stimuli was withheld, than when access to less

[12] Due to the correlative nature of this evidence it is not clear as to whether these events served any motivational function. Getting up late for example, may have been correlated with other events (such as an increased rate of demands, restricted access to tangibles) that actually functioned as EOs.

preferred stimuli was withheld. Interestingly aggression still occurred in conditions in which highly preferred items were freely available and only less preferred items were restricted. This may reflect within-session changes in preference during the course of a functional analysis condition or may be a discriminative effect. Alternatively it may be that restriction per se acts on the value of tangible items.

Consequences that are provided in a functional analysis for members of an alternative response class may act as an MO for tangible maintained challenging behaviour. Asmus, Franzese, Conroy, and Dozier (2003) provided some evidence to suggest that the consequences provided for stereotypical behaviour during a functional analysis served as an AO for the tangible-maintained destructive behaviour of one individual. Although this was not replicated, when stereotypy was ignored in a subsequent functional analysis then tangible-maintained destructive behaviour occurred at higher rates than had previously been the case. Such findings draw attention to the problems associated with including multiple topographies of challenging behaviour in a functional analysis.

Six studies were identified that examined the role of the onset of demands as an EO for tangible-maintained challenging behaviour (Adelinis & Hagopian, 1999; Fisher, Adelinis, Thompson, Worsdell, & Zarcone, 1998; Fritz, DeLeon, & Lazarchick, 2004; Hagopian, Bruzek, Bowman, & Jennett, 2007; Murphy, Macdonald, Hall, & Oliver, 2000; Wilder, Chen, Atwell, Pritchard, & Weinstein, 2006). These studies suggest that in some cases, challenging behaviour may occur in the presence of demands, not necessarily because the demand is aversive per se, but because the demand interrupts ongoing free operant behaviour and the removal of the demand allows for the reinstatement of the preferred activity. Hagopian et al (2007) found that the challenging behaviour of three individuals that occurred in demand contexts was occasioned by a request that led to the termination of a preferred activity with which the participant had been engaged. A prior functional analysis for all three participants had failed to evoke similar levels of challenging behaviour as that which occurred in their natural environment. Subsequent analysis for one participant showed that challenging behaviour was not only evoked by 'do' requests (i.e., when the child was asked to complete a task) but also 'don't' requests (i.e., when the child was told directly not to engage in the activity). Thus high rates of challenging behaviour occurring in the demand condition of a functional analysis may at times be indicative of a tangible function *either* as opposed to *or* as well as an escape function. It would seem possible that such a situation could over time lead to the demand acquiring aversive properties by functioning as a reflexive CEO. For example, repeated pairing of a previously neutral demand with the subsequent unavailability of certain preferred items, would lead to the demand itself acquiring evocative properties independent of the availability of preferred tangibles. That is, the onset of the demand would signal the subsequent 'worsening' of the individual's condition (in the form of interruptions to ongoing activities) and would establish its own offset as an effective source of reinforcement.

The past decade has seen considerable advancement in our understanding of the influence of MOs on tangible-maintained challenging behaviour. A handful of studies have gone beyond within-session manipulations, to examine the influence of pre-session access to tangibles as well as other variables, on tangible-maintained challenging behaviour. Parameters surrounding the implementation of the tangible condition in a functional analysis have also been examined. For example, consequences provided for alternative response classes and the relative preference for tangible items have all been shown to influence tangible-maintained challenging behaviour. Studies have also highlighted the role that the onset of demands may have as a MO for tangible-maintained challenging behaviour.

Social-Negative Reinforcement

Epidemiological studies suggest that, in some 34.2% of cases, challenging behaviour may be maintained by the contingent removal of aversive stimuli, such as academic demands (Hanley, Iwata, & McCord, 2003). From an MO perspective, the onset of an aversive stimulus establishes its own offset as an effective form of reinforcement and evokes negatively reinforced behaviour (J. Michael, 1982, 1993). Such MOs may have a *phylogenic* basis and function as a UMO, for example as with the onset of pain, or may have an *ontogenic* basis and function as a type of CMO, whereby a previously neutral stimulus has acquired similar value- and behaviour-altering properties to that of the UMO or other CMO with which it has been associated.

A decade ago understanding of the influence of MOs for negatively reinforced challenging behaviour was, in comparison to other functions, relatively well developed. In his paper McGill argued that the onset and offset of demands functioned as an MO for challenging behaviour maintained by negative reinforcement. A number of studies that had examined such relations were cited (e.g., Vollmer, Marcus, & Ringdahl, 1995). Several studies had by this stage begun to investigate the specific stimulus properties of demands that functioned as MOs for negatively reinforced challenging behaviour, such as the rate of demand presentation (Smith, Iwata, Goh, & Shore, 1995), difficulty of demands (Weeks & Gaylord-Ross, 1981) and the novelty of demands (Mace, Browder, & Lin, 1987). Other studies had demonstrated the *abolishing* and *abative* effects of certain antecedent manipulations, such as providing advanced notice of transition (Tustin, 1995) or embedding demands in preferred activities (Carr, Newsom, & Binkoff, 1976). It was also acknowledged that other stimulus changes, such as the onset of social attention (Taylor & Carr, 1992) or ambient noise (O'Reilly, 1997), could evoke negatively reinforced challenging behaviour. Thus relatively idiosyncratic stimulus parameters had been identified as MOs for negatively reinforced challenging behaviour.

Research over the past decade on MOs for negatively reinforced challenging behaviour has progressed along these same lines. The current review identified 31 studies published from 1998-2007 to have investigated MOs for negatively reinforced challenging behaviour (see Appendix 3).

Investigators have continued to demonstrate that the onset of demands within a functional analysis may evoke escape maintained challenging behaviour (e.g., Roane, Lerman, Kelley, & Van Camp, 1999). That is, the onset of a demand acts as an EO establishing its own removal as an effective type of reinforcement.

Specific stimulus parameters of demands appear to act as an MO for escape-maintained challenging behaviour and the extent to which these parameters evoke challenging behaviour varies across individuals. The following variables have been manipulated within a demand context and been shown to influence escape-maintained challenging behaviour; choice over the task (McComas, Hoch, Paone, & El-Roy, 2000; Romaniuk et al., 2002), task familiarity (Asmus et al., 1999), use of an instructional strategy (McComas, Hoch, Paone, & El-Roy, 2000), noise (O'Reilly, Lacey, & Lancioni, 2000), task preference (Boelter et al., 2007), task repetition (McComas, Hoch, Paone, & El-Roy, 2000), presence of preferred stimuli (Long, Hagopian, DeLeon, Marhefka, & Resau, 2005) and the type and timing of prompting procedures (Borrero, Vollmer, & Borrero, 2004; Crockett & Hagopian, 2006; Ebanks & Fisher, 2003; Peyton, Lindauer, & Richman, 2005). These studies have built on the line of research exemplified by Smith, Iwata, Goh and Shore (1995) to demonstrate that relatively idiosyncratic aspects of academic demands can influence the occurrence of escape-maintained challenging behaviour. In order to effectively incorporate such variables it would seem important that indirect and/or descriptive assessments be conducted prior to a functional analysis (e.g., Borrero, Vollmer, & Borrero, 2004), so that the MOs that operate in the individual's natural environment can be successfully included in the demand condition of the functional analysis.

Michael (1993) distinguished between the surrogate and reflexive CEO in relation to negatively reinforced behaviour. The onset of a surrogate CEO establishes the effectiveness of the removal of some other stimulus event as an effective type of reinforcement, whereas a reflexive CEO relation establishes its own removal, as opposed to that of another stimulus as an effective source of reinforcement. Although a subtle distinction, it may be important to determine whether such relations apply to escape-maintained challenging behaviour. Conceptually, it appears that they may. For example, for some individuals the presence of specific stimuli, such as the use of certain prompts, appears to act as a type of *surrogate CEO*, in that it alters the aversiveness of the demand and thereby establishes escape from the demand as an effective source of reinforcement. In such a situation one would expect that not only is challenging behaviour higher in those conditions in which the prompt is present than when not, but also that task completion (presumably an indicator of the aversiveness of the task) is also differentially influenced by the presence of the prompt (e.g., Crockett & Hagopian, 2006). This type of relation certainly appears analogous to Michael's description of the surrogate CEO relation. Prompts may also act as a type of reflexive CEO (i.e., their onset may establish their own off-set as an effective reinforcer). In such a situation one would expect higher rates of challenging behaviour in conditions in which the prompt is present than when not, however one would not expect task completion (if it represents an indicator of the aversiveness of the demand itself) to be influenced (e.g., Peyton, Lindauer, & Richman, 2005). One would also expect challenging behaviour to maintain in conditions in which the prompt remains but the demand is no longer removed contingent on the behaviour (Peyton, Lindauer, & Richman, 2005).

The probability of escape-maintained behaviour may also vary as a function of the individual presenting the demand (Magito-McLaughlin & Carr, 2005; McAdam, DiCesare, Murphy, & Marshall, 2004). In a recent study, Magito-McLaughlin and Carr (2005) investigated the influence of 'rapport' on escape-maintained challenging behaviour[13]. Following a descriptive assessment staff members were assigned either to good 'rapport' or poor 'rapport' groups for each of three individuals with intellectual and developmental disabilities. For all three individuals challenging behaviour occurred exclusively in demand contexts, and was much more likely to occur in the poor 'rapport' dyads. Task completion was high in the good 'rapport' dyads but extremely low in the poor 'rapport' dyads. A multi-component intervention was introduced designed to improve the quality of 'rapport' between the poor 'rapport' dyads. Results showed that the intervention led to an increase in task completion and reduction in the likelihood of escape-maintained challenging behaviour. Thus, the nature of the person introducing a demand, and specifically how they go about doing it, may influence the occurrence of escape-maintained challenging behaviour.

Interestingly, some studies have gone beyond the examination of the onset of single antecedents (e.g., a demand) to begin to examine the influence that a combination of antecedent events may have on escape-maintained challenging behaviour (Call, Wacker, Ringdahl, & Boelter, 2005; Call, Wacker, Ringdahl, Cooper-Brown, & Boelter, 2004; Carey & Halle, 2002). Call et al (2004) for example reported that the escape-maintained challenging behaviour of three individuals only occurred in situations in which attention was not available. When non-contingent attention was provided during demand sessions, escape-maintained challenging behaviour occurred at relatively low

[13] 'Rapport' is likely to describe more specific dimensions of the interactions. For example, 'rapport' may simply describe a reinforcement history involving a high number of aversive events or a low number of reinforcing events.

levels. In a subsequent study Call (2005) found, for one individual, high rates of challenging behaviour occurring in the demand condition of a functional analysis only when access to tangibles was also restricted. Interestingly, neither the standard demand condition nor tangible condition consistently evoked challenging behaviour. These studies suggest that alternative sources of reinforcement may have an influence on the probability of escape-maintained behaviour occurring during demands. That is, providing non-contingent access to certain forms of positive reinforcement, such as attention or tangibles, may serve to abolish escape from demands as an effective type of reinforcement and thereby abate escape-maintained challenging behaviour. It seems important that descriptive assessments be conducted prior to beginning a functional analysis in order to identify such potential idiosyncratic relations.

The majority of studies to have investigated MOs for negatively reinforced challenging behaviour have relied on the systematic manipulation of within- as opposed to pre-session variables. However, pre-session variables are likely to influence the occurrence of such behaviour. For example Carr, McLaughlin, Giacobbe-Grieco and Smith (2003) reported that pre-session ratings of 'mood' predicted the subsequent occurrence of escape-maintained challenging behaviour[14]. Using a reversal design, Ducharme and Rushford (2001) found that child compliance with demands made by a brain-injured parent increased following pre-session play conditions. Such findings may have implications for the sequencing of functional analysis conditions. For example, the findings above suggest that for some individuals escape-maintained challenging behaviour may be less likely to occur following the play condition than following other conditions. Relations between pre-session events and negatively reinforced challenging behaviour clearly merit further investigation and appear to have been a relatively understudied area. One would expect that pre-session influences are likely to influence

[14] 'Mood' may simply reflect the inability to tact certain environmental events.

escape-maintained challenging behaviour. Failure to control for such influences would represent a threat to the internal validity of the functional analysis being conducted. For this reason it would seem important that such influences be monitored and where possible experimentally controlled.

There have been a number of studies to have investigated the influence of neurobiological variables, such as *naltrexone* (Garcia & Smith, 1999)[15] and *amphetamine* (Kelley, Fisher, Lomas, & Sanders, 2006) and physiological variables, such as menses (Carr, Smith, Giacin, Whelan, & Pancari, 2003), and sleep deprivation (O'Reilly & Lancioni, 2000) on escape-maintained challenging behaviour. For example, Kelley (2006) investigated the influence of amphetamine on the response allocation of a young boy with intellectual and developmental disabilities and ADHD between challenging behaviour and compliance within a demand context. When administered a placebo drug the boy allocated a greater percentage of responses to challenging behaviour than compliance. When amphetamine was administered there was a higher proportion of responses allocated to compliance. The influence of other medications, such as *risperidone* have also been investigated but without clear effects (e.g., Zarcone et al., 2004). Physiological factors may also influence escape-maintained challenging behaviour. O'Reilly and Lancioni (2000) reported that missing afternoon naps increased the occurrence of a young girl's escape-maintained challenging behaviour. Interestingly, it was also found that sleep deprivation influenced within-class response allocation, with aggression more likely to occur on days with an afternoon nap and self-injury more likely to occur on days without a nap. Thus escape-maintained challenging behaviour is clearly influenced by the biological context in which it occurs. As well as demonstrating the influence of biological variables on escape-maintained challenging behaviour, both

[15] An alternative account for the effects of naltrexone in this instance is that it serves to establish a punishment contingency for challenging behaviours.

of these studies included response allocation as a dependent variable, suggesting that the MO may independently influence response allocation. Further research should begin to attend to the influence of the MO on response allocation.

Other studies have investigated the onset of stimulus events, other than demands, that may act as EOs for escape-maintained challenging behaviour. For example, the onset of attention (Hagopian, Wilson, & Wilder, 2001; Moore & Edwards, 2003; Moore, Edwards, Wilczynski, & Olmi, 2001), social proximity (Oliver, Oxener, Hearn, & Hall, 2001), ambient noise (Buckley & Newchok, 2006; McCord, Iwata, Galensky, Ellingson, & Thomson, 2001), specific food textures (Patel, Piazza, Layer, Coleman, & Swartzwelder, 2005), and transitions (McCord, Thomson, & Iwata, 2001) have all been shown to evoke negatively reinforced challenging behaviour.

This body of research holds important implications for the design of conditions within a functional analysis (see Table 2.2). The play condition is designed to provide an analogue of the 'enriched' environment and represents a control condition. If high rates of challenging behaviour occur in this condition a judgment regarding behavioural function may be difficult to make. However, the play condition may contain potential EOs for negatively reinforced challenging behaviour, such as high levels of social attention, noise, social proximity. High levels of challenging behaviour occurring in the play condition of a functional analysis should prompt further investigation and it may be necessary to implement a modified condition in order to test whether a particular aspect of the play condition, such as social attention, evokes negatively reinforced challenging behaviour (cf., Hagopian, Wilson, & Wilder, 2001).

As already stated, several studies have examined the specific properties of demands that function as EOs for negatively reinforced challenging behaviour. Similar studies have now begun to be conducted for other sub-categories of negatively reinforced challenging behaviour. For example, Buckley and Newchock (2006) conducted a component analysis which revealed that the specific source of music was the critical variable that operated as an MO for challenging behaviour maintained by the removal of music. That is, high rates of challenging behaviour occurred when music was played on a tape but low rates when the same music was played via a CD. Likewise, Patel et al (2005) examined the specific textures of food that were associated with food packing, with higher levels of food packing occurring in conditions that involved higher textured foods. One possible account for this may be that higher textured foods require a greater response effort than do lower textured foods and as such are more aversive (Patel, Piazza, Layer, Coleman, & Swartzwelder, 2005). It seems important that research continues to investigate such stimulus parameters for all categories of negatively reinforced challenging behaviour.

The specific parameters of demands that exert value- and behaviour-altering effects over negatively reinforced challenging behaviour have continued to form a predominant theme in the MO literature. However, there have been several notable developments over the last 10 years. Studies have begun to examine the influence that combined antecedents may exert on negatively reinforced challenging behaviour. A handful of studies have investigated the influence of pre-session manipulations on negatively reinforced challenging behaviour, although further research along these lines is required. A host of biological factors have now been shown to influence escape-maintained challenging behaviour. Perhaps the most significant development has been in the investigation of negatively reinforced challenging behaviour evoked by stimuli other than demands, such as the onset of attention. This may have important implications for the design of specific functional analysis conditions and should prompt further investigation.

Automatic Reinforcement

In some 15.8 % of cases, challenging behaviour occurs independent of its social consequences and is automatically reinforced (Hanley, Iwata, & McCord, 2003). Challenging behaviour may at times be maintained by automatic negative reinforcement, such as the removal or attenuation of pain (DeLissovoy, 1963; Symons, 2002), however such relations have rarely been examined in the literature. As such the current section shall focus exclusively on evidence for the influence of MOs on challenging behaviour maintained by automatic positive reinforcement.

In his review McGill (1999) discussed research that indicated a role for MOs in automatically reinforced challenging behaviour. The deprivation of environmental stimulation was cited as a likely EO for automatically reinforced challenging behaviour (e.g., Berkson & Mason, 1964). Likewise the non-contingent provision of environmental stimulation was recognised as most likely operating as an AO for such behaviours (e.g., Kennedy & Souza, 1995). One study was also cited which suggested that providing access to stimulation that was matched to that which maintained challenging behaviour was more effective in reducing such behaviours than providing access to unmatched stimuli (Piazza et al., 1998). Fluctuations in specific, as opposed to general, sources of stimulation may function as an MO for automatically reinforced challenging behaviour.

There have been some notable developments over the past decade in our understanding of the variables that exert a *motivative* influence over automatically reinforced challenging behaviour, as well as in the methods used to investigate such variables. The current review identified some 36 studies carried out from 1998-2007 which involved the assessment of MOs for automatically reinforced challenging behaviour (see Appendix 4).

It has been suggested that *response deprivation* and *satiation* may function as an MO for challenging behaviour (Klatt & Morris, 2001). That is, the restricted opportunity to engage in a free operant behaviour may establish engagement in that behaviour as an effective source of reinforcement and evoke associated responses. Likewise, the value of the consequences that maintain a response that is forced above its free-operant baseline rate will abolish and associated behaviours will abate. There is some evidence to suggest that such relations may influence automatically reinforced challenging behaviour (Rapp, 2004, 2006; Rapp, Vollmer, St Peter, Dozier, & Cotnoir, 2004). For example Rapp (2004) found that the stereotypy of a young boy with intellectual and developmental disabilities occurred at lower rates after having had prior access to the response earlier in the day; in contrast Rapp (2006) found that one individual's stereotypy increased immediately following a response blocking intervention. Thus the opportunity to engage in automatically reinforced challenging behaviour may function as an MO for future occurrences of the same response.

A number of studies have demonstrated that providing access to competing sources of stimulation, that are not necessarily matched to that which maintains challenging behaviour, may act as an AO for automatically reinforced challenging behaviour (e.g., Cuvo, May, & Post, 2001; Healey, Ahearn, Graff, & Libby, 2001; Long, Hagopian, DeLeon, Marhefka, & Resau, 2005; O'Reilly, Murray, Lancioni, Sigafoos, & Lacey, 2003; Rapp, Vollmer, St Peter, Dozier, & Cotnoir, 2004; Roane, Kelly, & Fisher, 2003; Roscoe, Iwata, & Goh, 1998; Wilder, Kellum, & James, 2000). O'Reilly et al (2003) found, for example, that providing non-contingent access to vibrating stimuli reduced the attempted self-injury and agitation of a man with intellectual and developmental disabilities that appeared to be automatically reinforced.

One concern associated with providing non-contingent access to competing sources of stimulation for automatically reinforced challenging behaviour is that with repeated exposure the value of the unmatched stimulus is abolished; whilst leaving intact the EO for challenging behaviour. One would expect therefore that over time challenging behaviour would re-emerge as the reinforcing value of the unmatched stimuli is abolished. In their study, Lindberg, Iwata, Roscoe, Worsdell, and Hanley (2003) found that increasing the length of experimental sessions to 120 mins, led to the re-emergence of automatically reinforced self-injury in two individuals, when providing access to unmatched stimuli had previously been effective in reducing self-injury during brief experimental sessions. A number of parameters may be important in ensuring that competing sources of stimulation maintain their effectiveness over time, such as ensuring the alternative stimuli are preferred (Ahearn, Clark, DeBar, & Florentino, 2005), providing access to multiple as opposed to single sets of stimuli (DeLeon, Anders, Rodriguez-Catter, & Neidert, 2000; Lindberg, Iwata, Roscoe, Worsdell, & Hanley, 2003) and combining the provision of unmatched stimuli with other procedures such as response blocking (Carr, Dozier, Patel, Adams, & Martin, 2002). Thus providing non-contingent access to competing sources of stimulation may act as an AO for automatically-reinforced challenging behaviour, however there are concerns regarding the extent to which such interventions can successfully maintain over time without additional procedures.

Some studies have compared the effectiveness of providing access to matched against unmatched sources of stimulation for automatically reinforced challenging behaviour with conflicting results. The hypothesis here is that providing access to matched sources of stimulation is more likely to act as an AO for challenging behaviour. Six studies were identified which found that providing access to matched stimuli led to greater reductions in automatically reinforced challenging behaviour than did providing access to unmatched stimuli (Fisher, Lindauer, Alterson, & Thompson, 1998; Higbee, Chang, & Endicott, 2005; Piazza, Adelinis, Hanley, Goh, & Delia, 2000; Piazza et al., 1998; Rapp, 2007; Taylor, Hoch, & Weissman, 2005). In contrast, however, Ahearn et al (2005) reported that the automatically reinforced challenging behaviour of two children with intellectual and developmental disabilities showed greater reductions when they were given access to highly preferred unmatched rather than matched stimuli. Likewise Carr et al (2002) failed to find any benefits of matched over unmatched stimuli. Given the results of the Lindberg et al study it would seem important that such comparisons involve analyses over extended periods of time. One would expect that the effectiveness of matched sources of stimulation in reducing automatically reinforced challenging behaviour would maintain over extended periods of time, whereas the effectiveness of unmatched sources of stimulation would not.

Deprivation of certain specific, as opposed to general, forms of stimulation therefore appears to act as an EO for automatically reinforced challenging behaviour and the non-contingent provision of matched sources of stimulation acts as an AO for such behaviours. As the response-reinforcement relation is typically covert in challenging behaviour maintained by its automatic consequences (Vollmer, 1994), investigators have had to rely on indirect assessments in order to inform hypotheses regarding the source of stimulation that maintains challenging behaviour.

Several studies have derived hypotheses regarding the specific maintaining variable for automatically reinforced challenging behaviour from the likely sensory products of certain topographies of challenging behaviour and provided access to matched sources of stimuli accordingly (Ahearn, Clark, DeBar, & Florentino, 2005; Carr, Dozier, Patel, Adams, & Martin, 2002; Fisher, Lindauer, Alterson, & Thompson, 1998; Kenzer & Wallace, 2007; Piazza, Adelinis, Hanley, Goh, & Delia, 2000; Piazza et al., 1998; Sidener, Carr, & Firth, 2005; Thompson, Fisher, Piazza, & Kuhn, 1998). Thompson Fisher, Piazza, and Kuhn (1998), for example, hypothesised that the automatically reinforced chin-grinding of a child with intellectual and developmental disabilities was maintained, specifically, by tactile stimulation to the chin and found that providing access to a source of stimulation matched to this hypothesis was effective in reducing the occurrence of chin-grinding. Deriving clear hypotheses from the topography of challenging behaviour may not always be possible however, particularly in cases where there are several potential sensory products to a response.

As such other studies have based such hypotheses on the results of indirect assessments, such as sensory extinction procedures (Rapp, Dozier, Carr, Patel, & Enloe, 2000; Rapp, Miltenberger, Galensky, Ellingson, & Long, 1999; Tang, Patterson, & Kennedy, 2003), antecedent 'sensory class' assessments (Britton, Carr, Landaburu, & Romick, 2002; Patel, Carr, Kim, Robles, & Eastridge, 2000; Taylor et al., 2005), and preference assessments (Goh, Iwata, & Kahng, 1999; Patel, Carr, Kim, Robles, & Eastridge, 2000; B. A. Taylor, Hoch, & Weissman, 2005). Rapp et al (1999) used a sensory extinction procedure in order to demonstrate that the hair pulling of a woman with intellectual and developmental disabilities was specifically reinforced by the digital-tactile stimulation it produced. Sensory extinction (i.e., the wearing of a rubber glove) led to a reduction in the amount of time spent manipulating hair. The non-contingent provision of hair that had been previously pulled out led to an increase in the amount of time spent manipulating hair and a notable reduction in hair pulling. Various types of preference assessments have also been used to help inform such hypotheses. Goh et al (1999), for example, conducted a preference assessment to identify the specific components of cigarettes that served to reinforce the pica behaviour of four individuals with intellectual and developmental disabilities. Other studies have relied on antecedent 'sensory class' assessments in order to identify potential matched sources of stimulation, for example Taylor et al (2005) reported that the automatically reinforced vocal stereotypy of one child occurred at lower rates in the presence of auditory toys than non-auditory toys. However, such indirect methods still do not allow for a definitive statement about whether a given source of stimulation is matched to or merely competes with that which maintains automatically reinforced challenging behaviour.

Given these problems investigators have begun to explore alternative methods to empirically identify sources of stimulation that are matched to that which maintains automatically reinforced challenging behaviour. Three studies were identified which utilised multiple-schedules in order to investigate sources of stimulation that may be matched to that which maintains automatically reinforced challenging behaviour (Rapp, 2006, 2007; Simmons, Smith, & Kliethermes, 2003). In line with the response deprivation hypothesis, if a source of stimulation provided during NCR matches that which maintains challenging behaviour one would expect there to be a decrease in the amount of time allocated to automatically reinforced behaviour after the intervention in comparison to before (i.e., due to reinforcer satiation). If however the stimulation competes with (rather than substitutes for) the maintaining variable then one would expect an increase in the amount of time allocated to the automatically reinforced response after the intervention. Rapp (2006) investigated the relative influence of providing non-contingent access to 'matched' stimulation and response blocking on the stereotypical behaviours of one individual. Relatively lower post-intervention levels of stereotypy were observed following non-contingent access to matched stimulation (suggesting it functioned as an AO), whereas there were relatively higher post-intervention levels of stereotypy following a response blocking component (suggesting it functioned as an EO). The results of both Simmons et al (2003) and Rapp (2007) also show consistent reductions in automatically reinforced challenging behaviour immediately following NCR involving 'matched' sources of stimulation. It would seem that the use of multiple-schedules may constitute an important methodological development in the investigation of MOs for automatically reinforced challenging behaviour. Future research is required that utilises such schedules in comparing the relative effectiveness of providing access to matched and unmatched sources of stimulation. One would expect there to be higher levels of automatically reinforced challenging behaviour in the period following access to unmatched than matched stimulation.

The alone condition of a functional analysis typically involves the deprivation of stimulation, which is hypothesised to act as an EO for automatically reinforced behaviour. However, in some cases it appears that this may not be the relevant EO for automatically reinforced challenging behaviour. Several studies have demonstrated that such behaviours may be evoked by the presence as opposed to the absence of certain stimuli (Carter, Devlin, Doggett, Harber, & Barr, 2004; Friman, 2000; Rapp, 2004, 2005; Van Camp et al., 2000). For example, Friman (2000) reported that the thumb sucking of a young boy with intellectual and developmental disabilities occurred only in the presence of a cloth and not in its absence. The automatic consequences of thumb-sucking were no more available in the presence of the cloth suggesting that it acted not as a discriminative stimulus but as an EO. Likewise, Van Camp et al (2000) found that the inclusion of a vibrating ball in the alone condition of a functional analysis led to an increase in automatically reinforced hand biting. A component analysis demonstrated that it was the vibrations made by the toy (as opposed to the sound or protrusions) that were associated with an increase in hand biting in the no-interaction conditions. Again the automatic consequences of hand-biting were no more available in the presence of the toy, suggesting that it may have exerted a motivative influence over challenging behaviour. Such findings appear to have important implications for the functional analysis of challenging behaviour (see Table 2.2). Low rates of challenging behaviour that occur in the alone condition may not necessarily indicate the absence of automatically reinforced challenging behaviour if the stimuli that occasion such behaviours are not present. Equally, high rates of challenging behaviour that occur in conditions in which the child has access to various stimuli (such as the attention, play and tangible conditions) may not necessarily be indicative of socially-maintained challenging behaviour. To protect against making such Type I or Type II errors it seems that indirect and descriptive assessments should be used to identify potential idiosyncratic sources of variability for automatically reinforced challenging behaviour. In addition within-session analyses have been shown to be useful in identifying an automatically reinforced function following an unclear initial functional analysis (Roane, Lerman, Kelley, & Van Camp, 1999).

Automatically reinforced challenging behaviour has also been shown to be influenced by certain biological variables, which may exert both value- and behaviour-altering effects (Carter, 2005; Garcia & Smith, 1999). Naltrexone is an opiate antagonist that has been shown to be relatively successful in reducing certain topographies of challenging behaviour in some individuals (Thompson, Hackenberg, Cerutti, Baker, & Axtell, 1994). Garcia and Smith (1999) reported that naltrexone successfully reduced the occurrence of automatically reinforced head-banging of one individual. It may that naltrexone acted as a MO by either abolishing the reinforcing value of the automatic consequences of head-banging or alternatively by establishing the punishing value of the response itself[16]. It seems important that there is further investigation of the influence of neurobiological variables on automatically reinforced challenging behaviour.

There have been significant developments in our understanding of automatically reinforced challenging behaviour over the past decade. The results of several studies suggest that the restricted opportunity to engage in automatically reinforced challenging behaviour acts as an EO for subsequent engagement in that behaviour. Providing access to unmatched sources of stimulation has also been shown to act as an AO for

[16] The extinction of the response-reinforcement contingency is another possible interpretation here.

automatically reinforced challenging behaviour. The literature has placed a particular emphasis on the identification of matched sources of stimulation for automatically reinforced challenging behaviour and the majority of developments have taken place along these lines. However, there have also been important developments in other areas, particularly in the recognition that the presence of idiosyncratic variables may evoke automatically reinforced challenging behaviour and that biological variables may also play an important role in altering the reinforcing value of automatically reinforced challenging behaviour.

The Behaviour-Altering Effect of the MO

Michael and colleagues argue that the value- and behaviour-altering effects of the MO, whilst often related are in fact independent of one another (e.g., Laraway, Snycerski, Michael, & Poling, 2003). Thus an operative contingency of reinforcement is not a *necessary* condition for the MO to influence behaviour. Given a sufficient level of water deprivation one would be expected to search for water in places where reinforcement has historically been absent and no discernable discriminative stimuli are present. Therefore, the behaviour-altering effect of the MO can be isolated by demonstrating changes in behaviour that result from MO manipulations under conditions of extinction. Such relations may hold important implications for our understanding of challenging behaviour.

A recent body of work has attempted to isolate the behaviour-altering effect of the MO for both attention- and tangible-maintained challenging behaviour (O'Reilly, Edrisinha, Sigafoos, Lancioni, & Andrews, 2006; O'Reilly, Edrisinha, Sigafoos, Lancioni, Cannella et al., 2007; O'Reilly, Edrisinha, Sigafoos, Lancioni, Machalicek et al., 2007; O'Reilly et al., 2006). O'Reilly, Sigafoos et al. (2006) demonstrated that manipulating pre-session access to tangibles and attention respectively, influenced the tangible-maintained challenging behaviour of one individual and the attention-maintained challenging behaviour of another in subsequent test conditions, with higher levels of challenging behaviour occurring after deprivation conditions than after satiation conditions. These effects were found both when challenging behaviour was reinforced in test conditions and more importantly when the behaviour was placed on extinction, thereby providing evidence for the behaviour-altering effect of the MO independent of its value-altering effect. Other evidence would seem to support these findings, for example, NCR has been shown to be effective in attenuating the side-effects of extinction (Vollmer et al., 1998). It seems likely that NCR achieves such effects by exerting an abative effect on challenging behaviour in the absence of any reinforcement contingency.

However, an alternative account for these findings may exist. Perhaps the *value-altering* effect remains intact even when reinforcing consequences are removed. That is, the higher rates of challenging behaviour in EO conditions may be the result of an extinction burst. In contrast in AO conditions the 'extinction' procedure may not genuinely constitute extinction, as the event no longer contingent on challenging behaviour is not currently operative as a reinforcer and as such there is no extinction burst. This second interpretation implies that the removal of the reinforcement contingency may fail to isolate the behaviour-altering effect of the MO. Examination of within-session patterns may more readily enable the identification of an extinction burst, and would enable each of these alternative accounts to be empirically examined.

Whilst promising, there are some studies which appear to contradict these recent findings on the behaviour-altering effect of the MO. Two studies have failed to evoke consistently high levels of challenging behaviour in conditions in which an EO has been operative but challenging behaviour has been placed on extinction (Potoczak, Carr, & Michael, 2007; Worsdell, Iwata, Conners, Kahng, & Thompson, 2000). For example, Potoczak et al (2007) found that the challenging behaviour of four individuals occurred at consistently high rates in demand conditions in which both an EO (demand) and escape contingency were present. AB functional analyses (Carr & Durand, 1985), in which an EO but no reinforcement contingency was present, produced much more variable and less differentiated patterns of responding. However, the fact that challenging behaviour still occurred in the absence of an escape-contingency would seem to provide some tentative evidence for the behaviour-altering effect of the MO for escape-maintained challenging behaviour, in isolation of its value-altering effect. There are important differences between these studies, which both relied on within-session MO manipulation and those conducted by O'Reilly and colleagues, which involved pre-session manipulations. Conceptually, it would seem likely that pre-session experimental manipulations have a greater power (i.e., are more likely to exert an influence on behaviour-environment relations) and thereby are likely to function as a more potent EO than within-session manipulations[17].

Michael and colleagues suggest that the behaviour-altering effect of the MO may occur 1) directly, 2) by interacting with discriminative stimuli or 3) by influencing behaviours maintained by conditioned reinforcers (Laraway, Snycerski, Michael, & Poling, 2003; Michael, 1982, 1993). O'Reilly, Edrisinha, Sigafoos, Lancioni, Machalicek et al (2007) provided some evidence that the behaviour-altering effect of the MO may occur directly in the absence of any discernable discriminative stimuli. Following the pre-session deprivation of attention, the attention-maintained behaviour of one individual was shown to occur when they were alone and no discernable discriminative stimuli for attention were present. It seems important that further

[17] For example, being deprived of attention for an hour would seem likely to exert a greater influence on attention-maintained behaviour, than would the within-session deprivation of attention within the attention condition of a functional analysis (O'Reilly, 1999).

research begin to explore the other mechanisms by which the MO exerts its behaviour-altering effect.

These findings on the behaviour-altering effect would appear to hold several implications for our understanding of challenging behaviour, as well as our interpretation of the results of a functional analysis. If an EO can evoke challenging behaviour in the absence of a S^d or a reinforcement contingency then it would seem possible at least that the behaviour-altering effect may precipitate challenging behaviour in conditions of a functional analysis that are seemingly irrelevant to the function served by challenging behaviour (see Table 2.2). For example, given a sufficient level of tangible deprivation, an individual could display high levels of tangible-maintained challenging behaviour in the demand, attention, or alone condition of a functional analysis even in the absence of a relevant reinforcement contingency or any discriminative stimuli. Such a possibility, whilst speculative, would constitute a significant threat to the internal validity of functional analysis methods and in some cases may lead to the adoption of behavioural interventions that would otherwise be contraindicated. An MO analysis helps to identify potential threats to the integrity of a functional analysis and it would seem that these threats can be minimised by relatively simple modifications made to the standard functional analysis conditions (see Table 2.2). Taking the example of tangible-maintained challenging behaviour, one could take steps (by controlling pre-session and within-session access to tangibles) to ensure that the EO for tangible-maintained challenging behaviour is only in place in the tangible condition of the functional analysis and not in other irrelevant conditions. Examination of within-session patterns of responding would also help to clarify potential ambiguities that may arise when conducting a functional analysis. Given recent developments in our understanding of the behaviour-altering influence of the MO, it seems critical to ensure that the EO for a given behavioural function is present only in its relevant functional analysis condition and not in alternative, irrelevant conditions.

Table 2.2.

Implications of the Behaviour-Altering Effect for Functional Analysis

Condition with high CB	Probable function	Possible function	Unlikely function	Resolution
Attention	Attention	Tangible, Automatic	Escape	Provide access to tangibles (pre-session/within-session), remove stimuli that may evoke automatically reinforced challenging behaviour. Examine within-session responding.
Tangible	Tangible	Attention, Automatic	Escape	Provide access to attention (pre-session/within-session), remove stimuli that may evoke automatically reinforced challenging behaviour. Examine within-session responding.
Demand	Escape	Tangible, Attention, Automatic		Provide access to tangibles/attention, remove stimuli that may evoke automatically reinforced challenging behaviour. Examine within session responding.
Alone	Automatic	Tangible, Attention,	Escape	Provide access to tangibles/attention (pre-session only)
Play		Automatic, Escape	Tangible Attention	Remove stimuli that may evoke automatically reinforced challenging behaviour. Test potential EOs for negatively reinforced challenging behaviour

Suggestions for Future Research

There has over the past decade been a tremendous development in our understanding of the conditions that establish or abolish the motivation for the variables which maintain challenging behaviour. There are several areas where future research is clearly required to further enhance current understanding of the influence of MOs on challenging behaviour.

Currently, research has, with few exceptions, focused on the influence of the MO on a single response. However MO manipulations may influence response allocation between a number of concurrent operants. It seems possible, at least, that the MO may interact with aspects of the response-consequence relationship that have already been shown to influence response allocation (e.g., Horner & Day, 1991). Only three studies were identified in the current review that examined the influence of the MO on response allocation (Kelley, Fisher, Lomas, & Sanders, 2006; O'Reilly, Edrisinha, Sigafoos, Lancioni, Cannella et al., 2007; O'Reilly & Lancioni, 2000). It would seem important that future research begin to identify the extent to which the MO influences response allocation.

Related to this, the primary dependent variable used in MO research has typically been frequency. However, it has been suggested that the MO exerts an influence on other aspects of behaviour, such as response latency, relative frequency, response magnitude and so on (Michael, 2007). The demonstration that the MO alters the magnitude of a response or its latency would hold important implications for our understanding of the MO itself as well as for the development of treatment strategies. As such, applied studies should begin to examine the influence of the MO across a range of measurable dimensions of challenging behaviour.

A focus of many studies to date has been on the single effect of a particular antecedent manipulation as either an EO or AO. However, it seems possible that a single antecedent manipulation may have multiple value- and behaviour-altering effects on an array of reinforcers and punishers across different response classes. For example, providing high levels of social contact may abolish the reinforcing value of attention (acting as an AO for attention-maintained behaviour), whilst at the same time establishing the reinforcing value of a preferred toy (acting as an EO for tangible-maintained behaviour). Indeed Berg et al (2000) demonstrated that providing pre-session access to attention in a free play condition reduced the probability of a child choosing to interact with her mother (acting as an AO for attention-maintained behaviour), and increased the probability of choosing to play with preferred toys (acting as an EO for tangibly-maintained behaviour). This relation reversed following pre-session alone conditions. Applied research on challenging behaviour has to date, however, failed to take account of the multiple effects of MOs on challenging behaviour (Michael, 2007). Further studies should begin to address such relations.

The MO is a continuous variable[18] and it would seem important that future research reflect this complexity. Three studies have been conducted that have involved parametric analyses of the MO in relation to preference or adaptive behaviour maintained by tangibles (Gottschalk, Libby, & Graff, 2000; Klatt, Sherman, & Sheldon, 2000; McAdam et al., 2005). In their study, Gottschalk et al manipulated pre-session access to food tangibles between deprivation, control and satiation conditions, demonstrating that deprivation resulted in increased preference and satiation resulted in decreased preference in comparison to control conditions. It would seem important that such parametric analyses begin to be applied to the study of MOs for challenging behaviour.

The repeated presentation of a reinforcer is, over time, likely to exert abolishing effects on its current reinforcing value and abate those behaviours it currently maintains. There may be certain situations in relation to challenging behaviour when we wish to mitigate such effects, for example if teaching a functionally equivalent response. In a recent study, North and Iwata (2005) investigated the effects of various procedures

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[18] Although there are situations in which its effects are relatively discrete. See Michael's (1982) discussion of transitive CEOs for example.

designed to mitigate the abolishing effects of repeated reinforcer presentation on tangible maintained adaptive behaviours. Providing pre-session choice was shown to mitigate AO effects for two participants and within-session variations in the items used as reinforcers was shown to mitigate AO effects for another participant. It seems that further such studies are required that begin to investigate the influence of such manipulations on challenging behaviour.

There are a diverse range of biological influences that have, over the past decade, been shown to influence the occurrence of challenging behaviour. For example, genetic, physiological and neurobiological variables have all been shown to influence challenging behaviour. The MO provides a means of incorporating such influences into the functional analysis of challenging behaviour whilst retaining a conceptually systematic approach to their analysis. Further research is required to continue the developments that have been made in the integration of the behavioural and biological sciences in relation to the investigation of challenging behaviour.

Functional analysis provides an important tool with which to assess challenging behaviour as it develops in young children (Kurtz et al., 2003; Richman & Lindauer, 2005). It seems likely that at both phylogenic and ontogenic level MOs play a crucial role in the development of challenging behaviour and further empirical investigation in this understudied area is clearly required. Furthering current understanding of the role of the MO in the development of challenging behaviour would hold important implications for refining approaches to the early intervention and prevention of challenging behaviour in young children at risk of developing challenging behaviour.

A decade ago, McGill (1999) discussed the relevance CMOs held for our understanding of challenging behaviour. Whilst there have been several studies to have investigated the effects of CMOs on the occurrence of adaptive behaviours (e.g., Kuhn, Lerman, Vorndran, & Addison, 2006; Rosales & Rehfeldt, 2007) there have been no studies to explicitly address such mechanisms in relation to challenging behaviour. As already discussed, such relations may be relatively prevalent in precipitating challenging behaviour and it seems important that they begin to be empirically examined. Such studies would need to control for fluctuations in the UMO in order to demonstrate the presence of a CEO relation. Investigation of the procedures by which McGill (1999) has suggested that CEOs can be addressed in treatment, such as the extinction of CEOs, also seems to be important for future research. For example, if the presentation of a demand acts as a transitive CEO for attention-maintained challenging behaviour then the CEO relation could be extinguished by providing non-contingent attention during demand presentation, and by extinguishing the relation between challenging behaviour and attention.

Central to applied behaviour analysis has been the development of 'theory' inductively derived from the basic data it describes (Skinner, 1950). Providing a conceptual system within which empirical findings can be effectively described is an important goal for applied behaviour analysis (Baer, Wolf, & Risley, 1968) and one that is more likely to further our understanding of functional relations than an approach that relies on the empirical findings alone. Investigators studying the influence of the MO should continue to rely on the conceptual system provided by Michael in order to describe the functional relations they uncover. Some elements of this system (such as the 'CMO'), have not been utilised on occasions when it would seem that they provide a more accurate description of the facts than do alternative terms. Future research should rely on the full conceptual system provided by Michael, as opposed to select parts, when relating findings to basic principles of behaviour.

The list above is by no means exhaustive and there may be other areas that warrant further investigation, such as the value- and behaviour-altering influence of derived relational responding (Dymond, Roche, Forsyth, Whelan, & Rhoden, 2007;

Whelan, Barnes-Holmes, & Dymond, 2006) and the further investigation of the mechanisms by which the MO exerts its influence. A number of studies also seem to require systematic replication and extension, for example those that have focused on the behaviour-altering effect of the MO (e.g., O'Reilly et al., 2006) and the 'priming' effect of brief pre-session access to social positive types of reinforcement (Roantree & Kennedy, 2006).

Concluding Comments

Over the past decade there have been considerable developments in the investigation of MOs and the role played by such variables in challenging behaviour. The terms used to describe motivative events have evolved, as have the methods used to investigate their effects. This endeavour has served to facilitate the incorporation of MOs into the functional analysis of challenging behaviour and as such has been beneficial in developing our understanding of challenging behaviour.

There are several areas where future research is needed in order to continue this advancement. Of particular relevance to the remainder of the thesis is the emphasis given to the interaction between genetic variables and behaviour-environment relations.

The developments that have taken place over the past decade provide a firm foundation on which such developments can be built. The close connections between basic and applied research encouraged by the advent of functional analysis (Mace, 1994), will continue to be of critical importance in the future investigation of the effects of the MO on challenging behaviour.

Chapter III. A Functional Analysis of the Early Development of Self-Injurious Behaviour. Incorporating Gene-Environment Interactions[19]

"It is a mistake to ask which traits are hereditary and which are learned. Similarly it is a mistake even to ask how much hereditary and environment contribute respectively to any specified pattern of development. The correct question, as always, is how development takes place, in detail, step by step through the causal chains found operating in a specific individual under study." (Bijou & Baer, 1967, p. 111)

[19] An adapted version of this chapter was published in the American Journal on Mental Retardation. Langthorne, P. & McGill, P. (2008). A functional analysis of the early development of self-injurious behavior: Incorporating gene-environment interactions. American Journal on Mental Retardation, 113, 403-417.

Chapter Overview

The conceptualisation and study of the development of self-injurious behaviour has to date been characterised by genetic and environmental determinism. In the current chapter it is argued that gene-environment interactions may play an important role in the development of such behaviours for some children with intellectual and developmental disabilities. It is argued that genetic events may alter an individual's susceptability to the environmental causes of such behaviours by: 1) altering the initial uncommitted behaviours displayed by the young child, 2) altering the child's ability to discriminate certain stimulus events, 3) enduringly altering the reinforcing (and punishing) value of certain behavioural consequences. The applied implications of this expanded approach are discussed and a number of suggestions for future research are made. It is suggested that research be conducted that examines the motivative role of genetic events.

Introduction

The previous chapter provided a systematic review of the concept of the MO as applied to the functional analysis challenging behaviour. In the current chapter the focus is narrowed to provide an analysis of the implications of gene-environment interactions (GxE) for models of the development of self-injurious behaviour.[20]

The analysis of the early development of self-injurious behaviour has to date reflected the wider distinction between 'nature' and 'nurture'. Despite the status of genetic factors as risk markers for the later development of self-injurious behaviour, a model that accounts for their influence on early behaviour-environment relations is lacking. In the current chapter it is argued that the investigation of GxE and other forms of gene-environment interplay could potentially enhance current approaches to the study of self-injury. A conceptual model of the early development of self-injurious behaviour based explicitly on such relations is presented. The model is consistent with the basic tenets of functional analysis. Implications for research and the assessment, treatment and prevention of self-injurious behaviour are discussed.

Self-injurious behaviour is one of the most challenging forms of behaviour displayed by individuals with intellectual and developmental disabilities. Self-injurious behaviour refers to behaviours, such as head-hitting or scratching, that people direct towards themselves and which results in tissue damage (Tate & Baroff, 1966). Such behaviours have a pervasive impact on the quality of life of the individual, exerting not only negative physical but also negative social consequences (Robertson et al., 2004), and therefore represent a significant barrier to child development. Given its devastating

[20] To maintain consistency with the existing literature the chapter primarily considers self-injurious behaviour. Although this has received less attention, the same relations (i.e., the differential reinforcement of early child behaviour) are likely to apply to other forms of challenging behaviour, such as aggression or destructive behaviours. As such, the thesis of the current chapter is likely to also apply to other forms of challenging behaviour.

impact, intervening before self-injurious behaviour becomes an established part of the child's repertoire holds many apparent benefits. The advantages of early intervention for children with autism have been well documented (McEachin, Smith, & Lovaas, 1993), however with only a few exceptions (e.g., Wacker et al., 1998), the same cannot be said of early intervention for self-injurious behaviour. In part, this may be due to a limited understanding of the factors that lead to the genesis of self-injurious behaviour (Symons, Sperry, Holditch-Davis, & Miles, 2003).

Existing conceptual models of the early development of self-injurious behaviour have focused almost exclusively on the role of environmental factors (Guess & Carr, 1991; Kennedy, 2002b; Richman, 2008). Guess and Carr (1991), for example, argued that initially automatically reinforced stereotypical behaviours are shaped into increasingly severe topographies of self-injurious behaviour, as they begin to contact socially and non-socially mediated contingencies of reinforcement. Whilst a growing literature exists to support this model (e.g., Oliver, Hall, & Murphy, 2005; Richman & Lindauer, 2005) the influence of biological and genetic factors on early behaviour-environment relations remains poorly understood.

GxE have been investigated and shown to exert powerful effects in the development of several physical and mental health disorders (see Rutter, Moffitt, & Caspi, 2006 for a review). The GxE approach has developed from the observations that many behavioural disorders are causally influenced by environmental pathogens but with wide individual differences in response to such pathogens (Caspi & Moffitt, 2006). In these fields it is now recognised that the traditional view of there being additive, and non-interactive effects between genes and environment is somewhat misplaced. However the distinction has to some extent remained intact in the investigation of self-injurious behaviour. The investigation of GxE is one component of a more general approach towards the study of gene-environment interplay; an approach which also comprises examination of the effects of environment on gene expression, the effects of environment on genetic heritability and correlations between genes and environment (Rutter et al., 2006). In what follows the consequences of maintaining the false dichotomy between 'nature' and 'nurture' are discussed, before an expanded model of the early development and maintenance of self-injurious behaviour is proposed. The model presents a functional analysis of self-injurious behaviour based explicitly upon GxE and other forms of gene-environment interplay.

Genes and Self-Injurious Behaviour

Self-injurious behaviour in certain cases is clearly influenced by genetic sources of variability. Evidence suggests that certain forms of self-injurious behaviour may constitute part of the behavioural phenotype of a number of genetic syndromes. Gene to behaviour associations of varying specificity have been repeatedly demonstrated across a number of syndromes; including Lesch-Nyhan syndrome (Nyhan, 1972), fragile X syndrome (Symons, Clark, Hatton, Skinner, & Bailey, 2003), and Smith-Magenis syndrome (Finucane, Dirrigl, & Simon, 2001).

Few would subscribe to the view that genes 'cause' such behaviours. There is considerable within-syndrome variability in the extent to which individuals with a given syndrome go on to develop behaviours considered 'phenotypic' (Hodapp & Dykens, 2001). Environmental factors have been shown to contribute to such variability (Hessl et al., 2001). Even in cases where strong gene-behaviour associations do exist it does not necessarily follow that these occur independent of environmental influence. For example, even at the molecular level the environment has been shown to alter gene expression. For example, Restivo et al (2005) showed that environmental enrichment was associated with a number of changes at the behavioural and neurological level in FMR1-knockout (FMR1-KO) mice. Indeed, exposure to enriched environments was shown to alter gene expression in the visual cortex for both FMR1-KO mice and a wild strain of mice. Environmental influences, such as smoking, have been shown to alter the extent to which genes are expressed in humans (Spira et al., 2004). Gene-behaviour associations reflect not only the direct effect of genes but also the effects of environment and, where present, the effects of gene-environment interplay. It is not necessarily the case, therefore, that a strong gene-behaviour association indicates the absence of environmental influence. Despite the apparent ubiquity of gene-environment interplay, however, most behavioural phenotype research has failed to go beyond the demonstration of simple gene-behaviour associations (Hodapp & Dykens, 2001).

The continued neglect of environmental influences in behavioural phenotype research may limit our understanding of the development of self-injurious behaviour and paradoxically the role that genes play in this process. As Moffitt, Caspi and Rutter (2005) state:

> *Ignoring nurture may have handicapped the field's ability to understand nature* (p.478).

The functional effects of genes upon behavioural development remain poorly understood. There is a need for behavioural phenotype researchers to go beyond gene-behaviour association and to begin to incorporate GxE relations and other forms of gene-environment interplay into the study of self-injurious behaviour.

Environment and Self-Injurious Behaviour

Others have focused exclusively on the environmental determinants of self-injurious behaviour. Functional analysis is the hallmark of the applied behaviour analytic approach to the assessment and treatment of self-injurious behaviour (Hanley, Iwata, & McCord, 2003). Over the past decade, investigators have begun to incorporate an individual's biological functioning into the analysis of such behaviours (Langthorne, McGill, & O'Reilly, 2007). This has had a profound impact on the assessment and treatment of self-injurious behaviour displayed by people with intellectual and developmental disabilities.

However, the influence of genetic and other biological variables has not yet been integrated with models of the early development of self-injurious behaviour. Despite the status of genetic syndromes as significant 'risk markers' for the later development of self-injurious behaviour (McClintock, Hall, & Oliver, 2003), a conceptual model that accounts for their influence on early behaviour-environment relations is lacking. Secondly, the focus of behaviour analysis on behavioural function has led to a neglect of form. As has been repeatedly demonstrated there are highly specific relationships between certain genetic syndromes and particular topographies of self-injurious behaviour which current 'operant' models say little about.

The omission of genetic influences from functional analysis stems from a 'misunderstanding' of the relations between biological and behavioural events and an assumption that such factors are private, inaccessible and in some cases hypothetical (Thompson, 2007). Such an omission is particularly striking given that central to the operant model, from which the functional analysis of self-injurious behaviour has itself evolved, is the phylogenic and ontogenic selection of behaviour (Skinner, 1966). Behaviour analysis as a philosophy and a science is contextual (Morris, 1988), and the occurrence of any response can only be understood in regard to the historical and current context (both genetic and environmental) in which that act is embedded. Paradoxically, despite the prominence Skinner gave to genetic influences, their analysis has remained largely outside the realm of applied behaviour analysis.

GxE Interactions and the Early Development of Self-Injurious Behaviour

A GxE approach is based on the assertion that environmental 'pathogens' cause behavioural disorders and genes influence susceptibility to these 'pathogens' (Caspi & Moffitt, 2006). Several studies have recently demonstrated that the effects of exposure to an environmental 'pathogen' may be conditional on a person's genotype. In the first such study, Caspi et al. (2002) demonstrated that a functional polymorphism in the gene encoding the neurotransmitter-metabolizing enzyme monoamineoxidase A (MAOA) served to moderate the effects of child maltreatment on the later development of anti-social behaviour. Specifically, individuals who had low levels of MAOA and experienced childhood maltreatment were more likely to develop anti-social behaviours than were individuals with a high-activity MAOA genotype exposed to the same 'pathogen'. Similar GxE relations have been shown to influence the development of psychosis in adolescent cannabis users (Caspi et al., 2005), and the development of ADHD symptoms (Brookes et al., 2006).

Genes do not code for specific behaviours, rather the effects of genes upon behaviour-environment relations are by virtue of their effects on the organism as a whole developmental system (Johnston & Edwards, 2002). This system comprises of bidirectional relations between environmental, behavioural, physiological, neural and genetic sources of variability (Gottlieb, 2003). The role of DNA is to specify the production of mRNA, which then in turn determines the production of the polypeptides that form proteins. It is these proteins that act upon the development of the individual. This process is epigenetic and is itself influenced by environmental factors. The role of genes therefore is to influence the development of the organism as a whole (across neural, physiological, and behavioural pathways); it is this whole organism which then interacts with the environment.

The model proposed in the current chapter is based on the thesis that in some cases genes influence susceptibility to known environmental 'pathogens' for the development of self-injurious behaviour. To date researchers have typically examined either genetic or environmental factors whilst overlooking the interaction between the two. A conceptual framework is needed to examine the influence of such interactions as they apply to self-injurious behaviour. The chapter provides an operant analysis of GxE and other forms of gene-environment interplay and suggest that genes may alter basic behaviour-environment relations by virtue of their effect on the developmental system as a whole. The implications of such a model for the study, treatment and prevention of self-injurious behaviour are discussed.

A Functional Analysis of the Early Development of Self-Injurious Behaviour

How do genes influence the development of the organism in a way that has specific effects on subsequent behaviour-environment relations? It has been postulated that genes may influence behaviour-environment relations in a number of ways (e.g., Moore, 2002; Skinner, 1966). Conceptually, at least, such factors may alter the developmental system in a way that influences; (1) the stream of 'uncommitted' behaviour from which an operant response evolves, that is they may contribute to initial behavioural variation. (2) the sensitivity of the individual to changes in environmental stimulation, that is they may either facilitate or inhibit the discrimination of stimulus events and (3) the value of certain environmental consequences that serve to reinforce or punish behaviour, that is they may establish or abolish the 'motivation' for the consequences that maintain self-injurious behaviour. These effects are likely to be achieved by the influence of genes on neurobiological and physiological pathways.

Figure 3.1. A model of the early development of self-injurious behaviour.

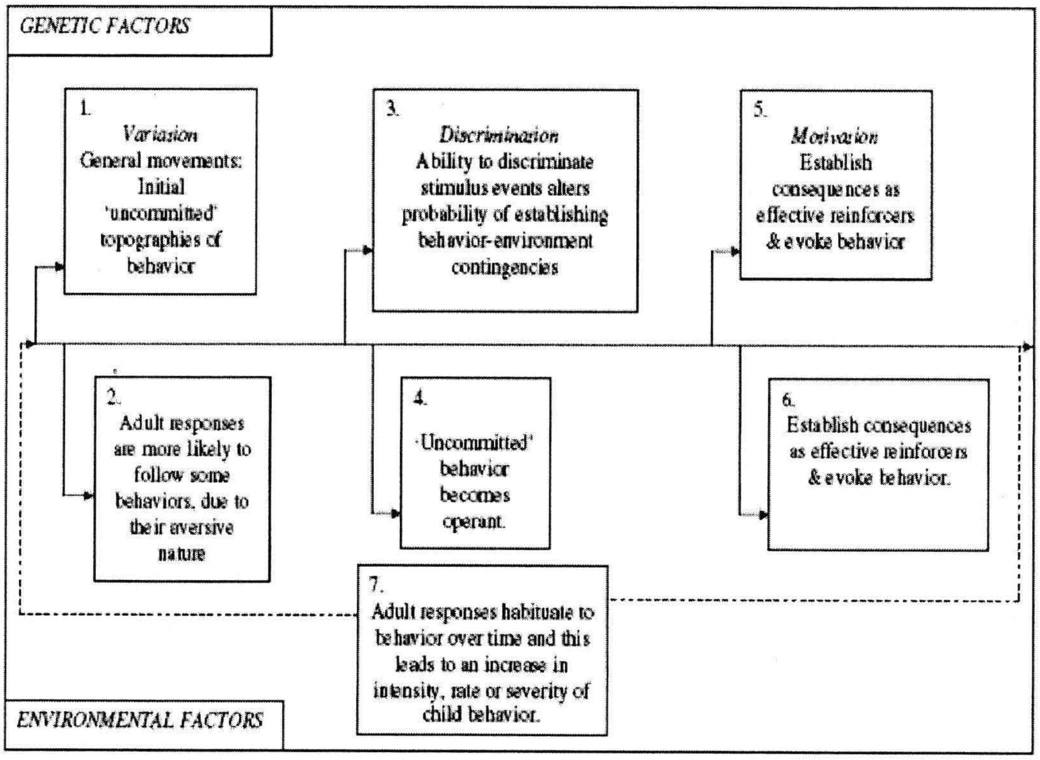


Figure 3.1 provides a schematic representation of a model of the early development of self-injurious behaviour based on the relations discussed above. Many of the environmental elements to this model have been comprehensively addressed in previous accounts of the development of self-injurious behaviour (Guess & Carr, 1991; Oliver, 1993). The influence of genetic factors and the role of certain other biological factors (such as health conditions), however, have to date escaped systematic appraisal. The model consists of seven stages, which for schematic purposes are presented in a linear fashion; this is not to imply that the model necessarily follows a linear path of causation or that all stages are necessary for the development of self-injurious behaviour.

In stage 1, genetic events alter the development of the individual in a way that influences the emission of 'uncommitted' topographies of behaviour from which an operant response evolves. The analysis of general movements may hold particular clues for our understanding of the later development of self-injurious behaviour (Symons, Sperry et al., 2003). Thus, genes contribute to initial behavioural variation, albeit pre and post-natal environmental factors may also influence this. In stage 2 it is recognized that some forms of uncommitted behaviour are more likely to elicit a social response than are others and this waxes and wanes over time as the environment itself adapts to the behaviour of the child. This stage is critical in the evolution from uncommitted behaviour to self-injurious behaviour. In stage 3 genetic events (in addition to pre- and post-natal environmental factors) alter individual development in such a way that determines the discrimination of stimuli. Thus genetic factors may in part determine the discrimination of certain stimulus events and thereby alter the likelihood with which certain contingencies are formed. In stage 4 self-injurious behaviour contacts socially and non-socially mediated contingencies of reinforcement to become operant. Both genetic (stage 5) and environmental (stage 6) events establish these contingencies as effective forms of reinforcement and evoke self-injurious behaviour by functioning as motivating operations. Finally in stage 7, the process of habituation shapes increasingly severe topographies of child behaviour.

Stage 1. Genetic Events Supply Uncommitted Topographies of Behaviour

A response must be first emitted 'for some other reason' before it can become operant. As Skinner noted:

> *Ontogenic contingencies remain ineffective until a response has occurred. In a familiar experimental arrangement, the rat must press the lever at least once 'for other reasons' before it presses it 'for food'* (1966 p. 1206).

It seems that genetic factors may play an important role in helping to determine the initial stream of 'uncommitted' behaviour, out of which increasingly complex operant responses evolve (MacLean, Ellis, Galbreath, Halpern, & Baumeister, 1991). In some cases such uncommitted behaviours may be of initial survival value to the organism, for example as in the rooting reflex of a newborn baby, and are the building blocks out of which complex behaviour, such as *manding* for food, are shaped. Evidence from animal models suggests that genetic manipulations influence the emission of motor activity (such as the degree of environmental 'exploration') which facilitates or inhibits the subsequent development of behaviour-environment relations (McKerchar, Zarcone, & Fowler, 2005). Both genetic and environmental factors alter the developmental system in such a way as to influence the initial uncommitted behaviours that an individual emits.

Newborn infants display prominent and complex movement patterns, termed general movements, which follow a predictable developmental course (Einspieler & Prechtl, 2005). Whilst general movements may be influenced by environmental factors, such as pre- or postnatal injury or exposure to toxins, there is a large genetic contribution to such movements. General movements have been observed both in vitro and post-natally and are thought to antecede the development of 'voluntary' (or rather operant) behaviour. Thelen (1979) noted the importance of early rhythmic motor behaviour for motor development in typically-developing infants. Qualitative differences in general movements have been shown to differentiate between low-risk and high-risk pre-term infants (Prechtl, 1990). In addition early patterns of general movements predict later problems in child development, such as cerebral palsy (Cioni et al., 1997).

It has been suggested that the analysis of general movements may hold some 'clues' for the later development of self-injurious behaviour (Symons, Sperry et al., 2003). However to date there have been no attempts to integrate this with existing conceptual accounts of self-injurious behaviour. The initial occurrence of general movements may represent the building blocks out of which self-injurious behaviour evolves. There are some parallels here with the motor control hypothesis of stereotypical and self-injurious behaviour. For example, evidence suggests that the motor control of individuals who display stereotypy differs from controls (Bodfish, Parker, Lewis, Sprague, & Newell, 2001). It has been suggested that the root of such differences in motor control may lie in a dopamine deficiency in basal ganglia functioning (Turner & Lewis, 2002). Work with deer mice has shown that the prevention or attenuation of stereotypical behaviours through environmental enrichment occurs only for mice who show enrichment-related differences in cortical-basal ganglia circuitry (Lewis, Tanimura, Lee, & Bodfish, 2007).

Rett syndrome and Down syndrome are two of the few genetic syndromes for which general movements have been empirically investigated (Einspieler, Kerr, & Prechtl, 2005; Mazzone, Mugno, & Mazzone, 2004). In a retrospective study, Einspieler et al. (2005) found that the quality of general movements in 26 infants, who had been diagnosed with Rett syndrome, was considerably impaired. By 4 months of age all of the infants had impaired 'fidgety' general movements, which were topographically different from those observed in infants with acquired brain lesions. Four of the infants with Rett syndrome were observed to have tremulous movements in the arms. The chromosomal basis of Rett syndrome lies in a mutation of the MeCP2 gene, a mutation which has pervasive effects on brain development. The disorder is associated with structural reductions in the basal ganglia and the decreased production of dopamine transporters, leading to the altered pattern of motor development associated with the syndrome (Schroeder et al., 2001). The effect of the MeCP2 gene mutation on brain development is likely to give rise to a pattern of impaired general movements that quickly contact contingencies of reinforcement. Indeed the stereotypical and self-injurious behaviours associated with Rett syndrome have been shown to be influenced by both automatic and socially mediated consequences (e.g., Oliver, Murphy, Crayton, & Corbett, 1993).

Such relations may hold relevance for the later development of self-injurious behaviour in other genetic syndromes. Lesch-Nyhan syndrome, for example, is characterized by a particular movement disorder (Nyhan, 1972). Early movements characteristic of this syndrome include involuntary spasmodic, wild, and violent movements of the face, shoulders and hips, which are present at 8-12 months of age. This precedes the development of self-injurious behaviour, which has a typical onset at between 2 and 3 ½ years of age. The movement disorder associated with Lesch-Nyhan syndrome has been associated with the impaired functioning of the dopaminergic system (Wong et al., 1996). The concentration of dopamine transporters does not, however, differentiate between individuals with Lesch-Nyhan syndrome who have both the movement disorder and self-injurious behaviour and those with only the movement disorder (Harris et al., 1999), suggesting that dopamine depletion is not sufficient to account for the development of self-injurious behaviour (Schroeder et al., 2001). There may be a GxE relationship underpinning this observation. For example, the HPRT deficiency results in changes in brain structure and function which leads to the emission of certain 'uncommitted' behaviours which are likely to expose a child with Lesch-Nyhan syndrome to environmental 'pathogens' for self-injurious behaviour. Hypothetically, a child with Lesch-Nyhan syndrome may accidentally bang their head as a result of reflex extension, part of the movement disorder associated with the syndrome, before that response comes under social control (Hall, Oliver, & Murphy, 2001). In the absence of differential social reinforcement (the environmental pathogen) SIB would not be expected to develop.

Smith-Magenis syndrome is a genetic syndrome associated with severe self-injurious behaviour, which includes the removal of finger- and toenails. Empirical evidence on the origin and function of this behaviour is scarce. However, Finucane et al (2001) speculate that its origin may lie in the altered neuropathy associated with the syndrome. Clinical signs of peripheral neuropathy have been reported in approximately 75% of SMS patients (Greenberg et al., 1996). Specifically, it has been hypothesized that abnormal sensations in peripheral nerves lead the child to pull at his or her toenails. Such 'uncommitted' behaviour is likely to quickly be selected by its environmental consequences (e.g., social contact from parents) and acquire operant status. Again, the hypothesis here is that the interstitial deletion of chromosome 17p11.2 in Smith-Magenis syndrome leads to damage of the peripheral nervous system, resulting in behaviour which exposes the child to known 'pathogens' for the development of self-injurious behaviour.

It has been argued that genes (in addition to pre- and post-natal environmental factors) influence the emission of uncommitted behaviours by acting on the individual at a neurobiological and physiological level. Specific examples of how and why this may occur have been provided for three different syndromes. It would seem that the assessment of general movements in children with genetic syndromes associated with self-injurious behaviour as well as the neurobiology underpinning such movements may be an important avenue for future research. This pattern of development, in which general movements precede the development of self-injurious behaviour, complements existing models and empirical evidence on the development of self-injurious behaviour.

Stage 2. The Environment is More Sensitive to Certain Topographies of Child Behaviour

Evidence suggests that one way in which genetic factors interact with the environment may be to indirectly influence the extent to which an individual is exposed to environmental 'pathogens' for a behavioural disorder (Rutter et al., 2006). Several studies have demonstrated the impact of such gene-environment correlations on the development of a range of disorders (Ge et al., 1996). In the context of the current discussion 'uncommitted' child behaviour, such as the reflex extension associated with Lesch-Nyhan syndrome, is likely to evoke a particular response from parents and caregivers. Once evoked these adult responses are likely to lead to the child being exposed to known environmental 'pathogens'.

It is the differential responsiveness of the environment to certain topographies of behaviour that is so critical for the environmental selection of operant behaviour (Bijou, 1966). To the extent that certain forms of child behaviour serve as aversive stimuli their onset may function as motivating operations (Laraway, Snycerski, Michael, & Poling, 2003) for the removal or attenuation of that behaviour. In the context of the current model parents and caregivers may be more likely to respond to particular 'uncommitted' behaviours (such as the wild and violent movements associated with Lesch-Nyhan syndrome) than they are to others. This is likely to be essential in the evolution from uncommitted behaviour to self-injurious behaviour. Parental responses that are successful in removing such behaviours are then more likely to occur in the future (Oliver, 1993). A broad range of contextual variables, such as parental stress (Hastings, 2002), may influence the degree to which parents are differentially responsive to atypical child motor movements.

There is a body of evidence to suggest that (a) particular topographies of child behaviour function as an aversive stimulus for parents and caregivers (e.g., Hastings, 2002), (b) child behaviour functions as a *motivating operation* for adult responses to that behaviour (e.g., Taylor & Carr, 1992) and (c) that the responses of adults serve to reinforce self-injurious behaviour (e.g., Richman & Lindauer, 2005). Thus adult responses to self-injurious behaviour are themselves shaped by the behaviour of the child. The role of genes in this process is to influence the likelihood of the child's exposure to such 'risky' environments.

Stage 3. Genetic Events Determine the Sensitivity of the Nervous System to Changes in Environmental Stimulation

Genetic events may help determine the sensitivity of the individual to various forms of environmental stimulation (Moore, 2002; Skinner, 1966). The ability to discriminate stimulus events is critical for the development of behaviour-environment relations. Someone who is congenitally blind is unlikely to be sensitive to a change in visual stimulation; conversely an individual with 20/20 vision is likely to be particularly sensitive to such a change. The result of such genetically (and environmentally) determined changes may be to enhance, or conversely diminish, an individual's susceptibility to the discrimination of certain stimulus events.

Some genetic influences may *restrict* the sensitivity of the individual to a stimulus event, whereas other genetic influences may *enhance* sensitivity to particular stimulus events. This is likely to be achieved by the influence of genes on the development of the individual at a neurobiological and physiological level. Reiss et al (2004) provided evidence that the impaired visual-spatial abilities and enhanced face processing and emotionality associated with Williams syndrome had specific neuroanatomical correlates. Magnetic resonance imagining scans showed that participants with Williams syndrome had reduced thalamic and occipital lobe grey matter volumes and reduced grey matter density in subcortical and cortical regions comprising the visual-spatial system, in comparison to controls. The Williams syndrome group also showed increases in volume and grey matter density in areas known to be involved in emotion and face processing. Thus, a specific genetic event (interstitial deletion of chromosome 7) was shown to be related to neuroanatomical changes, which themselves are related to the ability to discriminate specific stimuli. Clearly environmental factors may also contribute to such individual differences in brain development. Given that the ability to discriminate a stimulus change is a necessary pre-requisite for the establishment of behaviour-environment contingencies then such differences may hold an important function for the early development of self-injurious behaviour. It is surprising therefore, that such factors have not, to date, received attention in existing models of the early development of self-injurious behaviour.

A number of genetic syndromes may be associated with the altered ability to discriminate certain stimulus events. For example, individuals with fragile X syndrome (FXS) appear to be particularly sensitive to changes in social and sensory stimulation (Kau et al., 2004), perhaps potentiating the establishment of contingencies between their behaviour and social/sensory consequences. Hyperacusis (sensitivity to sound) is present in as many as 95% of individuals with Williams syndrome (Klein, Armstrong, Greer, & Brown, 1990). Genetic events may also reduce sensitivity to certain stimulus events. Cornelia de Lange syndrome, for example, is associated with minimal responsiveness to sound or pain (Johnson, Ekman, Freisen, Nyhan, & Shear, 1976). A reduced sensitivity to pain may prevent the establishment of a contingency between self-injurious behaviour and its painful automatic consequences.

Whilst the degree to which autism can be regarded to be a genetic disorder is of some controversy, what has been repeatedly demonstrated is that many individuals diagnosed with autism show an altered ability to discriminate certain stimuli. For example, autism is associated with an enhanced response and slower habituation rate to the repeated presentation of a tactile stimulus (Baranek & Berkson, 1994). Likewise, autism is associated with an enhanced auditory discrimination (O'Riordan & Passetti, 2006) whereas certain visual stimuli are less likely to be discriminated (Bertone, Mottron, Jelenic, & Faubert, 2003). Although the source of such variability (i.e., genetic or environmental) is of some debate, it does seem possible that the altered ability to discriminate changes in stimulation may influence the later development of behaviour-environment relations.

In sum, both genetic and environmental events may influence the extent to which changes in environmental stimulation are discriminated; a necessary pre-requisite for the development of an operant response, such as self-injurious behaviour. Such relations may alter the individual's responsiveness to known environmental 'pathogens' for the development of self-injurious behaviour, and thereby constitutes a potential GxE.

Stage 4. Child Behaviour Meets Contingencies of Reinforcement & Forms Part of a Response Class

A primary aim of functional analysis is to identify those consequences that maintain self-injurious behaviour. It is beyond the scope of this chapter to provide a review of this area; however, three areas of research are of particular relevance.

Firstly, research spanning three decades has shown that self-injurious behaviour displayed by people with intellectual and developmental disabilities may be evoked and maintained by its environmental antecedents and consequences (Hanley et al., 2003). Secondly, it has been demonstrated that self-injurious behaviour displayed by individuals with genetic syndromes may be influenced by environmental events. For example, Hall, Oliver and Murphy (2001) demonstrated that patterns of self-injurious behaviour displayed by three boys with Lesch-Nyhan syndrome were consistent with a positive reinforcement hypothesis. Such evidence demonstrates that self-injurious behaviours displayed by individuals with genetic syndromes may be influenced by environmental contingencies. Thirdly, empirical evidence shows that social and non-social reinforcement is critical in the early development of self-injurious behaviour (e.g., Oliver et al., 2005; Richman & Lindauer, 2005). In sum, a body of evidence exists to suggest environmental factors are critical in the development and maintenance of self-injurious behaviour, even when that behaviour is specifically associated with genetic syndromes. Such factors represent known 'pathogens' for the development of self-injurious behaviour and it may be that genetic variables influence the extent to which an individual exposed to such environmental events goes on to develop self-injurious behaviour.

Stage 5. Genetic Events Function as Motivating Operations

The variables of which behaviour is a function comprise of more than the contingency between response and consequence. One important contextual variable is the motivating operation. Motivating operations refer to stimuli, stimulus conditions or operations that alter the value of consequences as reinforcers and the probability of behaviours being evoked that have historically been associated with those consequences (Laraway et al., 2003). The analysis of such events has been typically restricted to environmental events, such as the deprivation of attention or onset of aversive stimuli (McGill, 1999). However, genetic events may also alter the value of those consequences that maintain self-injurious behaviour (Kennedy, Caruso, & Thompson, 2001). Evidence from animal models exists to suggest that specific genetic manipulations may exert motivative effects on the reinforcing value of certain forms of reinforcement (Couppis, Kennedy, & Stanwood, 2008; Hayward & Low, 2007; Thomsen & Caine, 2006).

Although the evidence-base is small and somewhat limited, individuals with certain genetic syndromes may be more likely to display behaviours that serve specific functions. A number of single-case studies have shown that stereotypical and self-injurious behaviours associated with Rett syndrome may be especially likely to be maintained by either escape from aversive stimuli and/or automatic reinforcement (Iwata, Pace, Willis, Gamache, & Hyman, 1986; Oliver et al., 1993; Roane, Piazza, Sgro, Volkert, & Anderson, 2001; Wales, Charman, & Mount, 2004; Wehmeyer, Bourland, & Ingram, 1993). Likewise, two studies have reported on socially avoidant behaviours, including self-injury, in Cornelia De Lange syndrome (Oliver et al., 2006; Richards, Moss, O'Farrell, Kaur, & Oliver, in press).

Prader-Willi syndrome (PWS) appears to be related to tangible-maintained challenging behaviours. A small number of studies have examined the relation between PWS and tangible-maintained behaviour. For example, Joseph, Egli, Koppekin, and Thompson (2002) reported people with PWS were more likely to wait for a longer delay in order to access a larger magnitude of food than were controls. Other studies have reported the effectiveness of reinforcing exercise with food for people with PWS (Caldwell, Taylor, & Bloom, 1986; Keefer, Jackson, & Pennypacker, 2000). Few have addressed the impact of PWS on the function served by challenging behaviour, although it would seem possible that the effects of this genetic event may serve as a MO for challenging behaviours maintained by access to food (Kennedy et al., 2001). Challenging behaviours in PWS may also serve additional functions, for example, Didden, Korzilius and Curfs (2007) examined the function of skin picking in PWS and reported that for 70% of individuals, skin picking served an automatic function. Woodcock, Oliver and Humphreys (2009) recently reported that over 80% of individuals with PWS were reported to display challenging behaviours following disruptions in routine or expectations.

Finally, studies which have relied on indirect functional assessment or antecedent manipulations suggest that FXS may be specifically associated with challenging behaviour, including self-injurious behaviour, that is maintained by the contingent removal of aversive stimuli (Hall, DeBernadis, & Reiss, 2006; Symons, Clark et al., 2003; Woodcock et al., 2009). A postal survey study conducted by Symons, Clark et al. (2003) found that parents reported 87% of participants with FXS displayed such behaviours in response to routine changes, and 65% in response to task demands. In contrast only 3% of participants were reported to display such behaviours to access attention. Hall et al (2006) recently reported a relatively large-scale study, involving some 114 children with FXS in which patterns of responding were directly observed during antecedent manipulation of environmental conditions. Challenging behaviours were more likely to occur in conditions characterized by high social or performance demands. Other studies have noted high levels of 'anxiety' in individuals with FXS in situations characterised by high social or performance-related demands (Hessl, Glaser, Dyer-Friedman, & Reiss, 2006; Lesniak-Karpiak, Mazzocco, & Ross, 2003).

Genes do not directly influence behaviour, and the relations discussed are likely to be influenced by the impact of genetic events upon the developmental system as a whole (Gottlieb, 2003). Some genetic syndromes are associated with certain physiological conditions, and it may be that such conditions play an important role in establishing the 'motivation' for self-injurious behaviour. For example, sleep disturbance is reported to be present in 65-100% cases of Smith-Magenis syndrome (Greenberg et al., 1996). In cases, in which sleep disturbance associated with Smith-Magenis syndrome is shown to influence self-injurious behaviour, then treatment of the sleep disturbance may be one possible early intervention. (De Leersnyder et al., 2007). Such relations may be important in regards to the function served by self-injurious behaviour associated with other genetic syndromes. For example, FXS is thought to be associated with the abnormal activation of the limbic-hypothalamic-pituitary-adrenal (L-HPA) axis, a system primarily involved in the human stress response, the activation of which leads to the secretion of cortisol (Hessl et al., 2002). Hessl et al (2002) found higher cortisol levels to be positively associated with parental reports of the severity of problem behaviours displayed by males and females with FXS. It may be that the absence of the FMR1 protein leads to changes in the development of the L-HPA axis which enduringly heightens the probability of negatively reinforced behaviour. In regards to PWS, Holland, Whittington and Hinton (2003) have suggested that the genetic abnormality underlying PWS leads to the impaired functioning of the hypothalamic pathways and the absence of metabolic and psychological changes that normally follow food intake. People with PWS have been shown to have an abnormally high blood level of the hormone ghrelin which serves to enduringly heighten the reinforcing value of food (Del Parigi et al., 2002).

In sum, self-injurious behaviour is unlikely to develop unless there exists some 'motivation' for the consequences that are responsible for its maintenance. It seems that genetic factors may in some cases be the source of such 'motivation'. Evidence for this remains at a preliminary stage and few studies have met the standards of an experimental functional analysis (Hanley et al., 2003). Further studies based on the group comparison of individual data sets from individuals with different genetic syndromes are required. In cases in which genetically based motivating operations are present it would seem likely that these effects are enduring, rather than momentary. In such cases, there may be a particular 'sensitivity' to small fluctuations in environmental levels of the reinforcer that maintains the behaviour. This is likely to form a GxE that potentiates known 'pathogens' for the development of self-injurious behaviour. Such influences are likely to have important implications for the early assessment and intervention of self-injurious behaviour and are not currently integrated with any existing conceptual model of the early development of self-injurious behaviour.

Stage 6. Ontogenic Factors (Physiological, Neurobiological and Environmental) Function as Motivating Operations

Ontogenic factors may also exert a momentary or more enduring influence on the reinforcing value of certain behavioural consequences by functioning as MOs. Environmental, neurobiological and physiological factors may all serve to establish the motivation for some of the consequences that maintain challenging behaviours (see Langthorne et al., 2007).

Environmental influences.

The environments in which many people with intellectual and developmental disabilities spend a large proportion of their time are replete with characteristics that establish the 'motivation' for self-injurious behaviour. Such behaviours may be more likely to develop in environments that are austere or barren of stimulation, lacking in attention and access to preferred items and activities, high in aversive stimuli and social control (e.g., Berg et al., 2000; O'Reilly, 1999). The implications of environmentally based MOs (challenging environments) have been comprehensively reviewed in the previous chapter. As such the following section places greater emphasis on neurobiological and physiological influences.

Neurobiological influences.

The interaction between neurobiological and environmental events has attracted increased attention in recent years (e.g., Jewett, Cleary, Levine, Schaal, & Thompson, 1995). The direct effects of some psychotropic drugs may function as MOs, by exerting value- and behaviour-altering effects. For example the stimulant drug *methylphenidate* (MPH) acts on dopaminergic neurotransmitters and has been used widely in the pharmacological treatment of behavioural problems associated with attention deficit hyperactive disorder (ADHD). MPH has been shown to alter the occurrence of challenging behaviour in specific environmental conditions (Dicesare, McAdam, Toner, & Varrell, 2005; Kelley, Fisher, Lomas, & Sanders, 2006; Mace et al., 2009; Northup et al., 1999; Northup, Fusilier, Swanson, Roane, & Borrero, 1997). Similar evidence exists for the opiate antagonist *naltrexone* (Garcia & Smith, 1999; Symons et al., 2001) as well as drugs whose effects are more gradual and less discrete such as *risperidone* (Yoo et al., 2003; Zarcone et al., 2004).

Psychotropic drugs may have a number of indirect and unintended effects. Such side effects may influence unintended classes of behaviour (Singh & Aman, 1990) and in some cases such side-effects function as MOs (Valdovinos & Kennedy, 2004). For example, a notable side effect of anti-epileptics is to increase levels of 'appetite' leading to increased food consumption and weight gain. This may have not only value-altering properties (i.e. establish food as an effective reinforcer) but also behaviour-altering effects (such as evoking aggression or self-injury that have led to access to food in the past).[21]

Neurological conditions, such as epilepsy, may have a similar effect. For example, Roberts, Yoder and Kennedy (2005) demonstrated that epileptic seizures experienced by three individuals were temporally associated with the occurrence of challenging behaviour as recorded by care staff. Such conditions could potentially exert a momentary motivative influence on challenging behaviour.

Physiological influences.

A child's physical and mental well-being may be directly related to the occurrence and non-occurrence of challenging behaviour (Carr & Smith, 1995).

A number of prevalence studies have suggested a link between psychiatric conditions and the presence of challenging behaviour (e.g., Reiss & Rojahn, 1993; Rojahn, Matson, Naglieri, & Mayville, 2004). Conceptually, at least, it appears plausible that psychiatric conditions may function in some contexts as MOs (Emerson, Moss, & Kiernan, 1999). For example, the onset of depression may have both establishing and abolishing effects. Depression is associated with a number of physical, cognitive and emotional symptoms, including fatigue, loss of energy, insomnia and hypersomnia. Such symptoms may serve as MOs for negatively reinforced behaviour; momentarily establishing the removal of a social demand as an effective type of

[21] See Roberts et al (2008) for a recent empirical examination of this hypothesis.

reinforcement and evoking those behaviours that have previously been negatively reinforced by its contingent removal.

Studies suggest a relatively high prevalence of sleep abnormalities in children with intellectual and developmental disabilities (Brylewski & Wiggs, 1999). Sleep deprivation may function as a MO for challenging behaviour displayed by children with intellectual and developmental disabilities (Horner, Day, & Day, 1997; Kennedy & Meyer, 1996; O'Reilly, 1995; O'Reilly & Lancioni, 2000). O'Reilly (1995) examined the interaction between sleep disruption and environmental factors in the aggressive behaviour displayed by a young man with intellectual and developmental disabilities. Aggression occurred at higher rates in demand conditions of a functional analysis when the young man had less than 5 hours sleep. Therefore the deprivation of sleep appeared to function as a MO for negatively reinforced aggressive behaviour.[22] Other physiological conditions, such as otitis media (O'Reilly, 1997), allergy symptoms (Carter, 2005; Kennedy & Meyer, 1996), menstrual discomfort (Carr, Smith, Giacin, Whelan, & Pancari, 2003), gastro-intestinal problems (Wacker et al., 1996), and intra-cranial pressure (Hartman, Gilles, McComas, Danov, & Symons, 2006) may exert a similar effect on challenging behaviour.

Prevalence studies have established that an association exists between the presence of physical and sensory impairments and challenging behaviour (Emerson et al., 2001). Conceptually at least such events may serve as MOs. For example, mobility and sensory impairments may restrict the extent to which an individual is able to independently navigate around their environment. In such circumstances access to certain classes of reinforcement, such as the provision of attention or preferred tangibles,

[22] See Kennedy (2002a) and Harvey et al (2004) for further experimental analysis of the association between sleep disruption and negatively reinforced behaviour.

may be restricted; establishing access to those stimuli as an effective form of reinforcement and evoking behaviour, such as aggression, that has led to this in the past.

In sum, a range of environmental, neurobiological and physiological influences have been shown to act as MOs for challenging behaviour. Children with intellectual and developmental disabilities may be at a heightened risk of experiencing such influences. It seems likely that these ontogenic factors may influence the development of self-injurious behaviours in such children.

Stage 7. Increasingly Severe Topographies of Self-Injurious Behaviour are Shaped Via a Process of Habituation

The final stage of the model functions as a feedback mechanism to account for the selection of increasingly severe topographies of self-injurious behaviour over time. Adult responses *habituate* to the repeated presentation of aversive stimuli, such as early forms of self-injurious behaviour. A number of empirical studies have shown that the repeated presentation of a reinforcer leads to habituation, which may bring about changes in operant responding (Murphy, McSweeney, Smith, & McComas, 2003).

For the parent, habituation may over time have an abolishing effect on the reinforcing value of reductions in early child self injury (Oliver, 1993). This may, for example, lead to the parent no longer providing attention or removing a demand contingent on the child's behaviour. For the child, this change in parent behaviour may lead to an extinction burst, characterised by changes to the topography of self-injury (e.g., an increase in the rate, intensity, or variability of the response). Such changes in child behaviour are likely to re-evoke the parental response that maintains child self-injury (e.g., the provision of attention or removal of demand). This final stage accounts for how initially uncommitted behaviours are transformed into increasingly severe topographies of self-injury.

Implications for the Study, Treatment, and Prevention of Self-Injurious Behaviour

In the current chapter existing models of the genesis of self-injurious behaviour have been extended by incorporating GxE relations and other forms of gene-environment interplay. It is argued that exposure to environmental 'pathogens' is necessary for the development of self-injury. An account of why certain genetic syndromes are a risk marker for the development of self-injury (McClintock et al., 2003) has been provided, suggesting that genes may alter susceptibility to such 'pathogens'. Specifically genes may influence the development of self-injurious behaviour by acting on the individual in a way that; (1) provides uncommitted behaviour out of which self-injurious behaviour later develops, (2) influences the discrimination of certain stimulus events (3) enduringly alters the value of certain consequences that function as reinforcers. However, self-injury would not be expected to develop in the absence of exposure to environmental 'pathogens'.

Such a conceptual expansion may hold important implications for the assessment, treatment and prevention of such behaviours. One of the benefits of this analysis is that it may enable the development of environmental modifications that provide a better fit to the needs of the individual. For example, knowledge of GxE relations and other forms of gene-environment interplay may facilitate environmental changes that serve to prevent or remediate self-injurious behaviour. In addition, adopting a more integrated perspective may have consequences for the study of self-injurious behaviour. Some of the applied and research implications of this expanded model shall therefore be discussed in greater detail.

Stage 1 suggests that uncommitted behaviours are the building blocks out of which self-injurious behaviour evolves. The comprehensive assessment of uncommitted behaviour may also hold implications for attempts at prevention. If a particular genetic syndrome is associated with general movements that are likely to evoke a caregiver response then a prevention-based approach, targeting parental responses to such behaviours may hold some promise in reducing the adventitious reinforcement of such behaviour. In addition, the absence of certain general movements could spur the development of a prosthetic environment, which facilitates the development of alternative behavioural sets.

As suggested in stage 3 of the model, genetic influences may also determine the discrimination of certain stimulus events. This may have a number of implications for the early prevention of self-injurious behaviour. Environmental or neurobiological modifications could be introduced to reduce the sensitivity of the individual to particular stimulus events. For example, earplugs could be offered to children with Williams syndrome to reduce the sensitivity of the individual to changes in noise levels (O'Reilly, Lacey, & Lancioni, 2000). In other cases environmental or neurobiological interventions may be required to improve the individual's ability to form certain behaviour-environment contingencies. For example, the use of the opiate antagonist *naltrexone* may enhance the development of a contingency between self-injurious behaviour and its automatic aversive consequences (Sandman, Barron, & Colman, 1990). This would have all the elements of a punishment procedure albeit with the aim of establishing a typical rather than atypical experience of pain.

In stage 5 it is suggested that genetic events may function as enduring motivating operations for self-injurious behaviour. This holds an array of implications for the treatment and prevention of self-injury. Early functionally equivalent responses could be reinforced using consequences for which 'susceptibility' is thought to exist at a time before a clear function for self-injurious behaviour is apparent. Such an approach would seem to be particularly necessary given the reported difficulty in ascribing function to self-injurious behaviour displayed by very young children (Kurtz et al., 2003). This area has been subject to only a small amount of research and a systematic approach is now required that develops our knowledge of the value- and behaviour-altering influences of certain genetic influences. Further investigation of the pathways underlying such relations may provide an additional point of intervention.

The enthusiasm surrounding gene-environment interplay has been dampened by legitimate concerns surrounding the pragmatic difficulties of conducting such research. In order for this conceptualisation to influence the behaviour of researchers these concerns need to be addressed. Moffitt et al (2005) have proposed a seven-step strategy for the identification of GxE interactions and, in comparison to other fields, investigators of self-injurious behaviour are in a relative position of strength to begin the search for GxE and other forms of gene-environment interplay. First, it is well established that there are certain syndromes associated with the heightened prevalence of self-injurious behaviour, but that there exists considerable within-syndrome variability. Second, there is clear empirical evidence of the causal status of environmental 'pathogens' for the development of self-injurious behaviour. Third, we have established methods of measuring the influence of such 'pathogens' over time. Fourth, behavioural phenotype research has led to the identification of several candidate 'susceptibility' genes for self-injurious behaviour. In short we know enough about environmental and genetic influences to begin to construct specific and testable hypotheses about relations between genes and the environment and their role in the development of self-injurious behaviour.

How can such interactions begin to be examined? Large scale, longitudinal, prospective cohort studies in which the individual acts as his or her own control are the most powerful means of testing specific GxE hypotheses (Moffitt et al., 2005). Studying self-injurious behaviour as it develops already forms an important research agenda (e.g., Richman & Lindauer, 2005); future studies could examine whether individuals with specific genetic syndromes differ in terms of their susceptibility to the known environmental 'pathogens' measured in such studies. Existing developmental studies could also be retrospectively examined to see whether differences exist in regards to the developmental pattern of individuals with certain genetic syndromes in comparison to others.

Specific hypotheses based on some of the postulations made in the current chapter would seem to warrant further examination. For example, the assessment of early movements displayed by infants with certain genetic syndromes, such as Lesch-Nyhan syndrome, may provide a fruitful source of research. The extent to which such differences predict the actual development of self-injurious behaviour, and whether the presence of environmental 'pathogens' are necessary for this to occur, would form an important research agenda. Further study of the potential motivative effects of genes upon behaviour-environment relations also seems to be an important research question. Group control studies that followed the methodology used in current behavioural phenotype research, except with an emphasis on function rather than form, may constitute one means of testing such hypotheses. In addition further investigation of the neurobiological and physiological pathways that underpin such relations would seem to be of considerable importance (Turner & Lewis, 2002). A focus on the proximal effects of genes on the developing organism (i.e., the endophenotype) may hold important implications for our understanding of gene-environment interplay (Gottesman & Gould, 2003). Animal models, which allow for a greater degree of experimental control, may provide an important tool in this quest, indeed empirical examples of gene-brain-behaviour-environment relations already exist in the experimental literature (e.g., Couppis et al., 2008; Hayward & Low, 2007).

In general, the analysis of GxE relations and other forms of gene-environment interplay has the potential to contribute to a more comprehensive approach to the understanding of self-injurious behaviour. Indeed if genetic effects operate through influencing susceptibility to environmental 'pathogens' then a reduction in such 'pathogens' will decrease the likelihood of the development of self-injurious behaviour. At a molar level this could contribute to effective environmental manipulations that prevent the development of self-injurious behaviour when an otherwise high risk for their development exists. Such an approach would be analogous to the prevention of the deleterious effects of phenylketonuria through dietary control.

Conclusion

The development of any operant response is embedded in a context of gene-environment interplay. As Gottlieb (2003) states:

> *There can be no genetic effects on behavior independent of the environment and there are probably no environmental effects on behavior independent of genetic activity (p. 351).*

This context is likely to involve mutual interactions between variables at the genetic, neurological, physiological, behavioural and environmental level (Johnston & Edwards, 2002). Such relations have not been integrated with existing models of the early development of self-injurious behaviour. In the current chapter a conceptual model of self-injurious behaviour has been provided, one that remains compatible with the basic tenets of functional analysis. GxE relations and other forms of gene-environment interplay may be critically important, particularly when self-injurious behaviour is correlated with genetic syndromes. The evidence reviewed suggests that genetic influences are an important source of variability; one that applied behaviour analysis may need to begin to come to terms with. The experimental tactic of demonstrating functional relationships between dependent and independent variables provides one means of uncovering the nature of this interaction.

Chapter IV.

Gene-Environment Interactions. An Indirect Examination

"Genetic variables may be assessed either by studying organisms upon which the environment has had little opportunity to act (because they are newborn or have been reared in a controlled environment) or by comparing groups subject to extensive, but on the average probably similar, environmental histories." (Skinner, 1966, p. 1209)

Chapter Overview

Gene-environment interactions (GxE) are likely to play an important role in the genesis of challenging behaviour, such as self-injury and aggression. The literature discussed in the previous two chapters suggests that one way in which genes influence the occurrence of challenging behaviour may be to enduringly alter the reinforcing value of the consequences that maintain such behaviours. The study reported in the current chapter aimed to provide an indirect examination of this question.

Both fragile X syndrome (FXS) and Smith-Magenis syndrome (SMS) are genetically determined causes of intellectual and developmental disability that are associated with a number of specific topographies of challenging behaviour. Previous work suggests that the function served by challenging behaviours in these groups may be very different. Using an indirect measure of behavioural function (*Questions About Behavioral Function scale;* Matson & Vollmer, 1995) the current study examined within- and between-group differences in challenging behaviour displayed by children with FXS and SMS, in comparison to a mixed etiology control group of children with intellectual and developmental disabilities. Both between- and within-group differences were found in the form and function of challenging behaviour. The implications of these findings for future research, as well as for the assessment and treatment of challenging behaviour are discussed.

Introduction

A number of researchers have suggested that one way in which genes influence the occurrence of challenging behaviour may be to enduringly alter the reinforcing value of the consequences that maintain such behaviours (Kennedy, Caruso, & Thompson, 2001; Langthorne & McGill, 2008; Oliver, 1993). For example, food appears to be enduringly established as an effective type of reinforcement for individuals with Prader-Willi syndrome (Holland, Whittington, & Hinton, 2003; Kennedy et al., 2001). If genetic events provide some of the 'motivation' for challenging behaviour then one would expect there to be differences (both between- and within-syndrome) in the function served by challenging behaviour across certain syndrome groups.

There is some evidence to suggest that such relations may apply to challenging behaviour in FXS and SMS. For example, evidence from recent studies suggests that individuals with FXS may be less likely to display challenging behaviour that is maintained by the provision of social attention and more likely to be maintained by the removal of aversive stimuli, and/or the provision of tangibles (Hall, DeBernadis, & Reiss, 2006; Symons, Clark, Hatton, Skinner, & Bailey, 2003; Woodcock, Oliver, & Humphreys, 2009). Symons et al for example, using an indirect measure of behavioural function reported that only 3% of children with FXS displayed attention-maintained self-injurious behaviour. Whereas 65-87% were reported to display self-injurious behaviour (SIB) in response to task demands and changes in routine. In contrast several clinical reports suggest that SMS may be associated with challenging behaviour that is particularly likely to be influenced by social attention (Dykens & Smith, 1998; Smith, Dykens, & Greenberg, 1998). A recent descriptive study by Taylor and Oliver (2008) reported that for four out of five children with SMS, challenging behaviour was more likely to occur following periods of low adult attention or following reduced levels of demands and was likely to lead to an increase in attention or demands.

The identification and delineation of behavioural phenotypes has gained considerable momentum over the last decade. During this time there have been several methodological developments that have taken place in this area. The following section describes and reviews some of the methodological issues that currently characterise behavioural phenotype research and how they relate to the design of the current study.

Experimental Design

The aim of behavioural phenotype research is to examine characteristic patterns of behaviour that are consistently associated with particular genetically-caused disorders. Researchers have adopted several different experimental designs in order to attempt to identify any such relationships.

Several studies have conducted within-syndrome comparisons in order to describe the 'phenotype' associated with a specific genetic syndrome. Sloneem (2005), for example, utilised a number of measures to describe the form and function of challenging behaviours displayed by a sample of 32 individuals with SMS. The study demonstrated that aggressive behaviours displayed by individuals with SMS frequently serve a communicative function. Such a strategy also holds particular benefits in identifying relative areas of within-syndrome strengths and weakness. For example, within-syndrome studies have demonstrated that individuals with FXS may have relative strengths in simultaneous information processing in comparison to sequential processing (Dykens, Hodapp, & Leckman, 1987). Whilst such studies hold considerable value in adding to existing knowledge of behavioural phenotypes, there are some notable disadvantages in relying upon within-syndrome comparisons. Primarily, this centres on the absence of an appropriate control group against which the performance of a target group can be compared.

Other approaches have involved between-group comparisons contrasting the performance of individuals with a specific syndrome against controls. Such an approach enables an assessment of whether the behaviours associated with a syndrome are different from what would 'typically' be expected. Agreement, however, about what constitutes 'typical' performance differs between studies (Hodapp & Dykens, 2001, 2004). Identifying an appropriate control group, against which the performance of the target group can be compared, is one of the most significant issues affecting between-group research in this field. Researchers have used several different types of control groups and each of these strategies has particular advantages and disadvantages.

Firstly, some studies have included a control group consisting of individuals without intellectual and developmental disabilities who are either of the same chronological age (CA) or mental age (MA) as the experimental group. Such an approach allows strengths or weaknesses in performance to be contrasted against typically developing controls. However, as Hodapp and Dykens (2004) note it is only comparisons with CA controls that can demonstrate whether certain behavioural repertoires are 'spared' in specific syndrome groups. For example, Majerus, Glaser, Van der Linden, and Eliez (2006) investigated verbal short term memory (STM) functioning in Velo-Cardio-Facial syndrome (VCFS) by comparing the performance of eight children with VCFS to CA matched controls on verbal STM tasks. The study demonstrated that retention of verbal item information was 'spared' in VCFS children with impairments specific to serial-order information. Whilst such studies are of use in identifying strengths and weaknesses for individuals with specific syndromes relative to MA or CA matched controls they do not necessarily demonstrate a behavioural phenotype. For example, it may be that differences in performance may be a function of having an intellectual and developmental disability *per se* as opposed to being related to a specific syndrome. For this reason researchers have tended to use control groups composed of individuals with intellectual and developmental disabilities.

There are several ways in which control groups composed of individuals with intellectual and developmental disabilities can be established. The first such strategy involves identifying a 'mixed etiology' group who are equated both for CA and MA. For example, Summers and Feldman (1999) investigated the behavioural functioning of 27 individuals with Angelman syndrome by comparing scores on the *Aberrant Behavior Checklist* to those of a mixed etiology group matched for MA and CA. The study demonstrated that individuals with Angelman syndrome had significantly lower scores on measures of irritability and lethargy. On face value at least this would appear to provide an appropriate comparison between those with and those without a specific syndrome. Hodapp and Dykens (2001, 2004) note several problems with this approach, however. Specifically, they note the difficulty in identifying a 'mixed etiology' sample that accurately reflects the population of people with intellectual and developmental disabilities, a process which is further hampered by a lack of epidemiological research and disagreements regarding the definition of intellectual disability.

In the final approach individuals with a specific syndrome are compared to specific groups either with or without intellectual and developmental disabilities. For example, Dykens, Leckman, and Cassidy (1996) compared the nature and severity of non-food obsessions and compulsions in 91 individuals with Prader-Willi syndrome (PWS) with a group of individuals with a diagnosis of OCD without intellectual and developmental disabilities. The PWS and OCD groups showed similar levels of symptom severity and numbers of compulsions; they also showed more areas of symptom similarity than difference. Other studies have sought to compare groups of individuals with specific syndromes to examine between-syndrome differences; some such studies have also included an additional 'mixed etiology' control group. Such studies enable researchers to examine the relative 'uniqueness' of behavioural phenotypes. Dykens and Kasari (1997) for example examined challenging behaviour displayed by 43 children with PWS in comparison to a group of children with Down syndrome and a 'mixed etiology' group of children with intellectual and developmental disability. This analysis demonstrated that 12 behaviours from the Child Behavior Checklist were significantly more likely to be reported for individuals with PWS in comparison to the other two groups.

In sum, a range of experimental designs have been used in order to investigate behavioural phenotypes associated with specific genetic syndromes. The strength of each specific design is a function of the particular research question, and certain designs are clearly not appropriate to answer certain research questions.

In the current study a between-group analysis was adopted, this involved an indirect comparison of the function of challenging behaviour between children with FXS and SMS and a 'mixed etiology' control group. This design allowed any differences in behavioural function between the two syndrome groups to be examined, whilst also enabling a judgement as to whether this was any different from what would typically be expected in the population of children with intellectual and developmental disabilities as a whole.

Participant Identification and Recruitment

The 'gold standard' for group-design studies requires the random assignment of participants to experimental conditions in order to control for extraneous subject variables. Due to the focus on etiology in behavioural phenotype research, however, participants cannot be randomly allocated to experimental groups; rather group assignment is determined by the presence or absence of a specific syndrome. This removes an element of experimental control and it is always possible for subject-variables other than the putative independent variable to exert a confounding influence. For example, environmental factors may contribute to between-group commonalities and differences.

One additional concern in behavioural phenotype research lies in the way participants are recruited. Convenience sampling is usually required in such research, especially when investigating a genetic syndrome with a relatively low prevalence such as SMS. Such sampling techniques may lead to the identification of samples of participants that do not accurately reflect the underlying target population. In behavioural phenotype research the most efficient form of recruiting participants is typically to contact the relevant parental support group for a particular syndrome. Parent groups have an interest in promoting research and are in a position to contact a relatively large number of families. This strategy holds several advantages to the researcher, and is especially efficient in terms of time and costs. Hodapp and Dykens (2004) note, however, that members of parental support groups may not be necessarily representative of underlying target populations. It is likely that such parent groups hold a greater proportion of white, English-speaking, middle-class and well- educated parents than would be expected in the underlying population.

Alternative methods of recruitment do exist which may help to overcome such problems. For example researchers can attempt to recruit participants through regional genetics centres or through special schools located in a specific area. Whilst such methods are likely to yield a more representative sample, they are considerably less efficient in terms of the financial and time costs involved. The primary means of participant recruitment in the current study was for this reason through convenience sampling.

Measuring Behavioural Phenotypes

A final concern for behavioural phenotype research lies in the methods used to measure behaviours associated with specific syndromes (Hodapp & Dykens, 2004). There are various *indirect* and *direct* methods, which can be utilised in order to measure behavioural phenotypes.

Indirect measurement tools rely on informants to provide information about behaviours associated with a specific syndrome. One of the benefits of indirect methods lies in their ease of use. Large numbers of participants can be used in such research in a relatively time-efficient manner.

Various indirect methods have been employed in behavioural phenotype studies. Some studies for example have used comprehensive checklists to identify behaviours associated with a specific syndrome. Finucane, Dirrigl and Simon (2001) developed a bespoke checklist to investigate self-injurious behaviours displayed by individuals with SMS. Using such scales enables the investigation of specific behaviours known to be associated with a specific syndrome. In cases where such behaviours are not already known, the use of such checklists may result in highly idiosyncratic behaviours (such as the *polyembolokomania* associated with SMS) being missed. Checklists are also of little use in identifying qualitative differences within- or between- syndrome groups or in the investigation of more complex repertoires of behaviour (Hodapp & Dykens, 2004).

Another common indirect method to have been used includes the use of standardised psychometric tools, such as the *Aberrant Behavior Checklist* (Aman, Singh, Stewart, & Field, 1985). The benefits of such scales are that they have demonstrated validity and reliability; they also allow a reference to norms, allowing an assessment of whether individuals with a specific syndrome show higher or lower levels of a specific behaviour than typically found.

There are some notable problems associated with indirect methods, however. Indirect tools measure what informants say about the behaviours of interest rather than the behaviours themselves. There is not necessarily a correspondence between these two things, which requires an inference to be made about the relationship between the event that was measured and the behaviour of interest. As Johnston and Pennypacker (1993) note:

> *It is incumbent on anyone who uses indirect measurement to provide scientific evidence that the data obtained from the measurement system precisely reflect in some way the subject matter about which conclusions are drawn. (p. 114).*

Although direct measurement places a greater demand on the researcher, it provides a more valid means of assessing the target behaviour of interest than do indirect measures. Direct measurement helps to ensure that *"the phenomenon that is the focus of the experiment is exactly the same as the phenomenon being measured" (Johnston & Pennypacker, 1993, p. 113).*

In the current study, which aimed to include a large number of participants, indirect methods were used to collect information on the sample. Indirect methods enabled an examination of the primary research question for a relatively large number of individuals.

In sum, the effects of genetic events on behavioural function has attracted increased attention in recent years. The current study aimed to extend this line of work by examining both between- and within-syndrome differences in the function served by challenging behaviour displayed by children with FXS and SMS, in comparison to one another and to a control group of children with intellectual and developmental disabilities of a mixed etiology. To the author's knowledge this is the first study to conduct such a between-group comparison using an indirect measure of behavioural function. Data were also collected on between-group differences in the form of adaptive and challenging behaviours.

Method

Experimental Design

A between-group comparison design was employed to examine the primary research question. Two experimental groups were used, parents of children with a confirmed diagnosis of FXS and parents of children with a confirmed diagnosis of SMS. A group of parents of children with intellectual and developmental disabilities of a mixed etiology were included as a control group.

Research Ethics

The current study received ethical approval from the Tizard Centre Ethics Committee. For participants from the SMS group recruited via Regional Genetics Centres ethical approval was also granted by a multi-site NHS Research Ethics Committee and the study also received local R&D approval from each individual NHS Trust who agreed to support the research.

Participants

All participants were parents of children with intellectual and developmental disabilities who displayed challenging behaviour. All children were between 5-21 years of age and had a confirmed diagnosis of FXS, SMS or intellectual and developmental disabilities of a mixed etiology. Allocation to groups was dependent on the genetic status of each participant's child or children. Parents of children with SMS and FXS were requested to provide confirmation of their child's genetic status or to provide consent for the investigator to request this information from the family's local Regional Genetics Centre where the initial diagnosis had been made or failing that from the family's paediatrician or GP. Independent verification of each child's genetic syndrome was received for all but six participants (4 participants with SMS and 2 participants with FXS). Some participants from the mixed etiology control group had a genetic syndrome other than SMS or FXS, although independent evidence of this was not requested. This was deemed acceptable as the aim of including a non-specific group was to provide a heterogeneous sample of individuals with intellectual and developmental disability.

Convenience sampling was used to recruit participants from each group. Participants from the FXS group and from the mixed etiology control group were contacted via the relevant parental support group. *The Fragile X Society*, a UK-based parent support group for families with a child or children with FXS agreed to contact families of children with a diagnosis of FXS who had previously indicated a willingness to take part in research. A total of 174 information packs were sent out. Consent forms were returned for a total of 46 participants of whom 34 took part in the study. Participants for the mixed etiology control groups were recruited through *The Challenging Behaviour Foundation*, a parent support group for families of children with intellectual and developmental disabilities who display challenging behaviour. Information packs and consent letters were sent to a randomly selected sample of 115 families from the *Challenging Behaviour Foundation* mailing list. A total of 35 individuals returned consent forms of whom 30 took part in the study.

Participants for the SMS group could not be recruited via the *Smith-Magenis Foundation*, a parent support group for families of children with SMS. The study received an unfavourable review from the Foundation's professional advisory group. The specific reasons as to why the study was rejected were never disclosed despite repeated requests and no opportunity was afforded to respond formally to any concerns. As such alternative methods of participant recruitment were pursued for this group. These included contacting potential participants via the *Challenging Behaviour Foundation* mailing list and newsletter, through the *Unique* (a parent support group for rare chromosome disorders) database, letters to all 52-week-a–year residential schools throughout the UK, letters to all special education schools in Kent, advertisements in the November 2007 *Contact a Family* newsletter and on the *Yahoo! SMS group.* In addition, a snowballing technique was adopted whereby all families who took part in the research were asked if they knew of anyone with a child with SMS who may be interested in taking part in the study. These methods led to a total of 18 participants with a child with SMS being identified. As such, NHS Regional Genetics Centres across the United Kingdom were then contacted with information about the study and were asked whether they would be willing to forward information about the study to families of children with SMS who had received services from their centre. Following ethical approval Regional Genetics Centres from the following NHS Trusts agreed to support the study and send out information to potential participants. The number of participants recruited via each NHS Trust is shown in brackets: Birmingham Women's Health Care NHS Trust (1), Belfast City Hospital Trust (2), Greater Glasgow-Yorkhill Division NHS Trust (1), Liverpool Women's Hospital NHS Trust (1), and Oxford Radcliffe Hospitals NHS Trust (1). This resulted in a total of 24 participants being included in the research who had a child with a diagnosis of SMS.

An initial power analysis using the conventions suggested by Cohen (1992) suggested that in order to achieve an acceptable level of power (0.8) each group would require a minimum of N = 21 for a large effect size (d = .4) at $\alpha = .05$. Participant recruitment continued up until sufficient N had been reached for each group or a statistical difference between the groups in the direction hypothesised had been found, whichever occurred first. There was a total of 34 participants in the FXS group, 24 in the SMS group and 30 in the mixed etiology control group. Descriptive statistics for participant characteristics are presented below in Table 4.1.

Table 4.1

Participant Characteristics

	FXS	SMS	Mixed Etiology
Chronological Age (months).			
Mean (SD)	133.5 (36.3)	141.4 (44.6)	121.3 (34.7)
Gender (%)			
Male	91.4	54.2	80
Female	8.6	45.8	20

There were no significant differences between the groups for chronological age. The one-way ANOVA showed F to be non-significant at the 0.05 level: $F(2, 85) = 1.195$; $p > 0.05$. Therefore the groups appeared to be matched for CA.

There were significant differences between the groups in the gender of participants. A chi square test of the null hypothesis that there were no gender differences between the groups showed significance beyond the .05 level: $\chi^2 (2) = 11.505$; exact $p = 0.003$.

Measurement Instruments

Participants were asked to complete the following measures: *Aberrant Behavior Checklist-Community version* (ABC-C; Aman, Burrow, & Wolford, 1995), the *Vineland Screener (Sparrow, 2000),* and the *Questions About Behavioral Function* scale *(QABF; Matson & Vollmer, 1995).* The *Self-Injurious Behavior* subscale of the *Behavior Problems Inventory* (Rojahn, Matson, Lott, Esbensen, & Smalls, 2001) was also used for those participants who had endorsed self-injurious behaviour items in the *ABC-C.* Each measure took approximately 15 mins to complete.

The *Aberrant Behavior Checklist (ABC)* was originally designed to measure challenging behaviour displayed by individuals in institutionalised settings. The *ABC* is an informant-based problem behaviour rating scale that was developed by factor analysis using a large number of individuals in institutional settings (Aman et al., 1985). The *ABC* consists of 58 items, each scored on a 4-point scale (0: not a problem, through to 3: problem is severe in degree). The items fall into five subscales: (1) *Irritability, Agitation, Crying* (15 items), (2) *Lethargy, Social Withdrawal* (16 items), (3) *Stereotypic Behavior* (7 items), (4) *Hyperactivity, Non-Compliance* (16 items), and (5) *Inappropriate Speech* (4 items). A number of psychometric studies have shown that the *ABC* is a reliable and valid behaviour rating instrument (see Aman, 2002). The *ABC* was modified for use with community-based individuals and the original factor structure of the *ABC* has been shown to be valid for the *ABC-C* (Aman et al., 1995).

The *Vineland Screener (VSC)* is a tool to assess domains of adaptive behaviour derived from the full Vineland and is intended for research purposes only. The *VSC* consists of 15 items in each of the following three domains: *Communication, Daily Living Skills*, and *Socialization*. There are also a smaller number of items for the *Motor Skills* domain. Respondents are asked several probe questions to explore their perception of the child's ability for each item. Items are scored on a Likert-type scale from 0-2; an N can also be scored if the individual does not have the opportunity to engage in certain behaviours. Age equivalent scores for each sub-domain can be derived from the Vineland Screener. A mean age equivalent score across the Communication, Daily Living Skills and Socialization subdomains has been used in prior research to have used the VSC in order to provide a global measure of intellectual functioning (e.g., Chadwick, Kusel, Cuddy, & Taylor, 2005; Charman, Howlin, Berry, & Prince, 2004). Sparrow (2000) reports that the *VSC* has high inter-rater reliability and each domain has high convergent validity with the respective domains on the full Vineland.

The *Questions About Behavioral Function* scale *(QABF)* is a 25-item questionnaire designed to identify the variables that maintain challenging behaviour. The QABF is scored on a 4 point Likert scale from never (0) to often (3). If an item does not apply then the N/A option may be endorsed. Its five subscales (*escape, attention, non-social, tangible, physical discomfort*) have been confirmed via factor analysis and the scale has good reliability (Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000). Paclawsky, Matson, Rush, Smalls, and Vollmer (2001) assessed the convergent validity of the *QABF*, reporting moderate agreement between the *QABF* and the *Motivation Assessment Scale* (Durand & Crimmins, 1988) and the *QABF* and the results of analogue assessments. Evidence also supports the predictive validity of the QABF. Matson, Bamburg, Cherry and Paclawskyj (1999) for example, demonstrated that the QABF successfully identified behavioural antecedents for 84% of individuals in their sample of 398 individuals with intellectual and developmental disabilities. Treatment based on a QABF-based functional assessment was more likely to lead to reductions in challenging behaviour than treatments that were not based on functional assessment.

Independent researchers have reported discrepant results for the psychometric properties of the QABF. Shogren and Rojahn (2003) failed to find any advantage of the QABF over the MAS, reporting comparable inter-rater reliability and test-retest reliability scores for both. Likewise Nicholson, Konstantinidi and Furniss (2006) reported reliability scores that were substantially lower than those reported by Paclawskyj et al. (2000).

The *Self-Injurious Behavior* subscale of the *Behavior Problems Inventory* (*BPI*) was also completed for those participants, who had endorsed self-injurious behaviour items on the *ABC-C*. The *BPI* has been found to be a reasonably reliable and valid measure (Rojahn et al., 2001). The Self-Injurious Behavior subscale consists of 14 items which are rated for frequency on a 5 point frequency scale (never to hourly).

Procedure

Participants for the FXS and mixed etiology sample were contacted via the *Fragile X Society* and the *Challenging Behavior Foundation* respectively. All parents were provided with an information sheet describing the aims of the study, a consent form, a copy of the *ABC-C* together with a glossary of terms and a pre-paid envelope. Each participant also received a covering letter from the relevant parental support group detailing their support for the research.

Potential participants from the SMS group were contacted via a number of different means (see above). The procedure above was followed for those parents who were recruited via parental support groups (*Challenging Behavior Foundation* and *Unique*) and Regional Genetics Centres. All other participants contacted the primary researcher independently. Once these participants had established contact then the procedure described above was followed.

All participants were asked to return a completed copy of the *ABC-C,* a completed consent form indicating a convenient time for the interviews to be conducted and for the FXS and SMS groups either confirmation of their child's genetic diagnosis or consent for the researchers to seek this information from relevant agencies. On receipt of this participants were then sent a confirmation of the scheduled interview time together with copies of the remaining measures that were to be completed over the phone. Parents were advised to have the questionnaires to hand for the telephone interview.

All phone calls began with the researcher introducing himself and establishing whether this was a convenient time for the interview. If necessary, interviews were rearranged for an alternative time. All interviews began with a brief description of the aims and structure of the interview. At the end of each measure, participants were offered the opportunity to take a break and resume the interview at a later date. For participants who had indicated in the *ABC-C* that their child displayed self-injurious behaviour the Self-Injurious Behavior subscale of the *BPI* was completed to gain more detailed information on the form of self-injury displayed by each child. On some occasions parents had not endorsed self-injurious behaviour items in the ABC-C when it would have been appropriate to do so. In these cases the BPI was also completed for these individuals. The *QABF* was then completed for each general topographical category of behaviour that had been rated as problematic in the *ABC-C* (aggression, self-injury, destructive behaviour). The *VSC* was then administered. The *VSC* consists of several age ranges (0-2yrs, 3-5yrs, 6-12yrs, and 13-18yrs). All participants were assigned to the age range immediately beneath their child's chronological age. For example, if their child was 15 years old then the participant was asked to complete the 6-12 yrs old age range. The appropriateness of this age range was then assessed, using the following protocol. If the participant achieved a score of 9 or 10 (max score = 10) after the first five items or 18 to 20 (max score = 20) after the first 10 items then a higher age range was selected. Conversely if the participant achieved scores of 0 to 1 after five items (min score = 0) or 2 or less after 10 items (min score = 0) then a lower age-range was selected. This process was repeated for each sub domain until an appropriate age range was selected.

At the end of the interview participants were thanked for their time and informed that they would be sent an indirect functional assessment summarising the responses made to the *QABF* within the next two weeks. Confirmation of the genetic status of each parent's child if not already provided was sought from Regional Genetics Centres, paediatricians or GPs. Participants from the FXS and SMS groups who were not recruited via NHS Regional Genetics Centres were also asked if they would be willing to consider their child participating in a subsequent study. All data were stored in a secure location and inputted into *SPSS version 15* for later analysis.

Results

Adaptive Behaviour

Groups were compared for levels of adaptive behaviour and independent functioning using the Vineland Screener. Age equivalent scores from the full Vineland manual were derived from the Vineland Screener equated raw scores. Table 4.2 presents descriptive statistics for each group.

The Kolmogorov-Smirnov test of goodness-of-fit was significant at the .05 level for the mixed etiology group on the Daily Living Skills sub-domain, suggesting that the data were not normally distributed. As such a non-parametric Kruskal-Wallis test was used to examine group differences across all sub-domains of the Vineland. To reduce the probability of making a Type I error by multiple comparisons, $\alpha = .05$ was divided by the number of comparisons made, resulting in a significance level of .012. Henceforth where the same process has been used to account for multiple comparisons, this shall be referred to as the Bonferroni adjustment. Mann Whitney tests were used to examine post hoc between-group differences using a significance level of 0.016 (Bonferroni adjustment).

Table 4.2

Vineland. Age Equivalent Scores (in months)

	FXS (N = 34)		SMS (N= 22)		Mixed Etiology (N=30)	
	M	SD	M	SD	M	SD
Overall Mean	46.9	21.2	56.0	18.9	42.3	23.6
Communication Skills	50.6	30.7	68.1	27.8	40.4	28.2
Daily Living Skills	42.1	26.2	50.9	19.6	42.77	30.4
Socialization	48.0	17.8	49.2	16.8	43.8	18.7

For overall Vineland mean age equivalent scores, the Kruskal-Wallis chi-square test was not significant at the 0.012 level: χ^2 (2) = 8.05; p = 0.018. The Kruskal-Wallis chi-square test was also not significant for the Daily Living Skills sub domain: χ^2 (2) = 5.727; p = .057; and the Socialization sub domain: χ^2 (2) = 1.205; p=.547.

There were significant between group differences for the Communication sub domain of the Vineland. The Kruskal-Wallis chi-square test was significant at the 0.012 level: χ^2 (2) = 12.99; p = 0.002. A series of Mann-Whitney tests showed: differences between children with FXS and SMS were significant at the .016 level (U = 203; p = .004), as were differences between the SMS group and mixed etiology group (U= 157.5; p = .001). Differences between children with FXS and the mixed etiology group were non-significant at the .016 level (U= 403; p = 0.149).

Challenging Behaviour. Form

The ABC-C and BPI-SIB subscale were used to assess group differences in the form and severity of challenging behaviour.

Aberrant Behavior Checklist.

Table 4.3 shows descriptive statistics for the between-group comparison for the overall and sub domain scores of the ABC-C.

Neither Levene's test nor the Kolmogorov-Smirnov test were significant at the .05 level for any group or sub-domain, suggesting that the data were normally distributed. As such a series of one-way ANOVAs were used to examine group differences on all sub domains of the ABC-C, using a significance level of 0.008 (Bonferroni adjustment).

Table 4.3

Aberrant Behavior Checklist. Descriptive statistics

ABC-C	FXS (N=35)		SMS (N= 23)		Mixed Etiology (N=30)	
	M	SD	M	SD	M	SD
Overall Score	65.9	35.3	74.1	28.9	77.0	28.9
Irritability	18.6	11.0	25.3	8.9	24.5	8.9
Lethargy	11.5	9.3	9.4	9.6	12.1	9.0
Stereotypical	8.1	5.2	6.8	4.5	8.9	5.8
Hyperactivity	21.2	11.4	27.0	9.6	26.9	13.2
Inappropriate Speech	6.5	3.4	6.0	3.1	4.8	3.9

There were no significant between-group differences on any sub domains of the ABC-C at the .008 level. The one-way ANOVA revealed no significant between-group differences on overall scores: $F (2, 85) = 1.065$; $p = 0.349$; the irritability subscale: $F (2, 85) = 4.344$; $p = .016$; the lethargy subscale: $F (2, 85) = .599$; $p = 0.551$; the stereotypical behaviour subscale: $F (2, 85) = 1.072$; $p = 0.347$; the hyperactivity subscale: $F (2, 85) = 2.564$; $p = 0.083$; or the inappropriate speech subscale: $F (2, 84) = 2.102$; $p = 0.129$.

Behaviour Problems Inventory. SIB subcale.

The BPI was completed for all participants who were reported to display self-injurious behaviour. Some 91.4% of the FXS group were reported to display some form of self-injurious behaviour, 95.8% of the SMS group and 80% of the mixed etiology group. Table 4.4 shows descriptive statistics for the total BPI SIB subscale scores.

Table 4.4

Behavior Problems Inventory. SIB subscale. Descriptive statistics.

BPI	FXS (N=32)		SMS (N=23)		Mixed Etiology (N=24)	
	M	SD	M	SD	M	SD
Total SIB Subscale	11.3	8.7	18.8	9.6	14.3	7.3

Neither Levene's test nor the Kolmogorov-Smirnov test were significant at the .05 level for any group on the BPI, suggesting the data were normally distributed. As such, a one-way ANOVA was used to examine group differences. The one-way ANOVA test revealed significant between group differences on the Total SIB subscale at the .05 level: $F(2, 76) = 5.156$; $p = 0.008$.

Bonferroni pair-wise comparisons revealed significant differences between the children with FXS and children with SMS ($p < 0.05$). There were neither significant differences between children with FXS and the mixed etiology group nor between the SMS group and mixed etiology group.

Challenging Behaviour- Function

FXS. Within-group analysis.

Table 4.5 and Figures 4.1-4.3 show descriptive statistics depicting the within-syndrome profile of scores on the QABF for children with FXS across self-injurious behaviour, aggression and destructive behaviours. There appears to be a similar differentiation between the five subscales across all topographies. Children from the FXS group appear to score lower on the attention subscale in comparison to other socially influenced subscales across all topographies. Scores on the demand and tangible subscales appear to be slightly elevated in comparison to all other topographies.

Table 4.5

Within-syndrome descriptive results of the QABF for children with FXS (Total Scores).

QABF Total Scores	Self-Injury (N=30)		Aggression (N=32)		Property Destruction (N= 23)	
	M	SD	M	SD	M	SD
Attention Subscale	3.20	3.77	3.65	3.99	2.78	3.46
Tangible Subscale	8.57	3.73	8.26	4.12	5.87	4.29
Demand Subscale	9.07	3.79	10.45	3.59	6.04	4.48
Physical discomfort Subscale	3.87	5.37	3.32	4.69	1.26	2.85
Automatic Subscale	6.13	4.29	1.23	2.23	5.65	5.31

The Kolmogorov-Smirnov test of goodness-of-fit was significant at the .05 level for at least one subscale across all three behavioural topographies, suggesting that the data were not normally distributed. As such a series of non-parametric Friedman tests were used to examine within group differences in behavioural function for all behavioural topographies, using a significance level of .016 (Bonferroni adjustment).

Figure 4.1. Boxplot for QABF total scores. Self-injurious behaviours

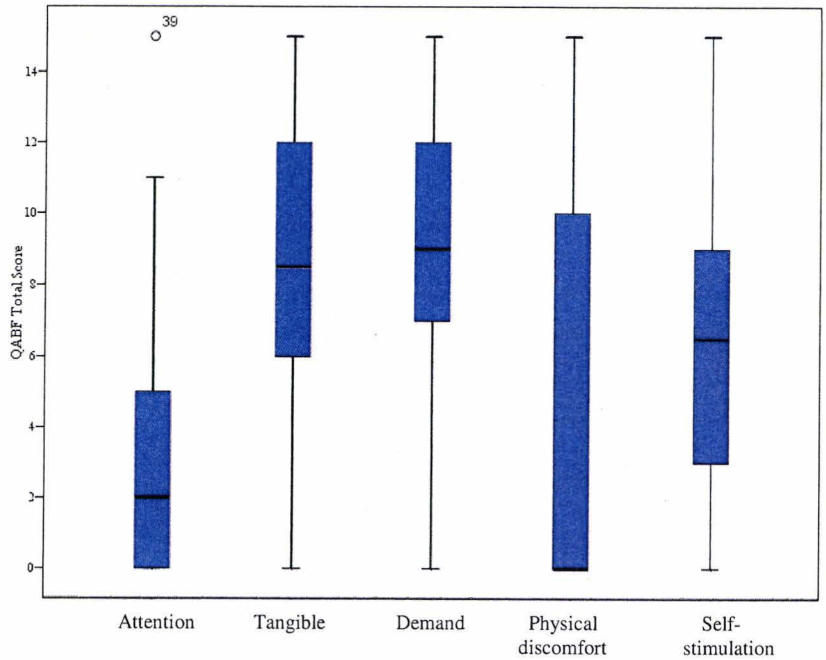


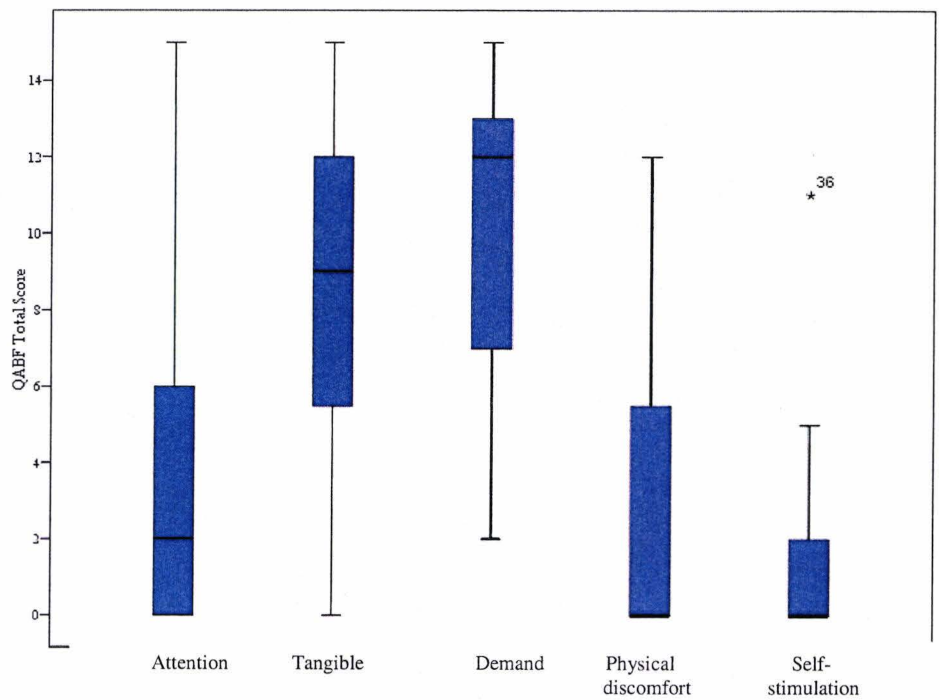
Table 4.6

Results of Pair-wise Comparisons on the QABF for Children with FXS who Display Self-Injurious Behaviour

Pair-wise comparison	Wilcoxon's W	N (excluding ties)	*p* (*sig at .005 level)
Attention vs. Tangible	27.5	28	.000*
Attention vs. Demand	29.5	29	.000*
Attention vs. Physical discomfort	147	25	.686
Attention vs. Self-stimulation	95	28	.012
Tangible vs. Demand	158.5	27	.471
Tangible vs. Physical discomfort	41.5	27	.000*
Tangible vs. Self-stimulation	90.5	28	.009
Demand vs. Physical discomfort	32	28	.000*
Demand vs. Self-stimulation	54.5	23	.009
Physical discomfort vs. Self-stimulation	115.5	27	.078

For self-injurious behaviour, the Friedman test was significant at the .016 level: χ^2 (4) = 35.88; p = 0.000. A series of Wilcoxon matched-pairs signed rank tests were conducted to compare pair-wise within group differences. The significance level of .005 (Bonferroni adjustment) was used for all non-parametric pair-wise comparisons. The results of each pair-wise comparisons are presented in Table 4.6 above. For self-injurious behaviours the following pair-wise differences were significant at the .005 level: attention<tangible; attention<demand; tangible>physical discomfort; demand>pain.

Figure 4.2. Boxplot for QABF total scores. Aggressive behaviours



For aggressive behaviours, the Friedman test was significant at the .016 level: χ^2 (4) = 69.856; p = 0.000.

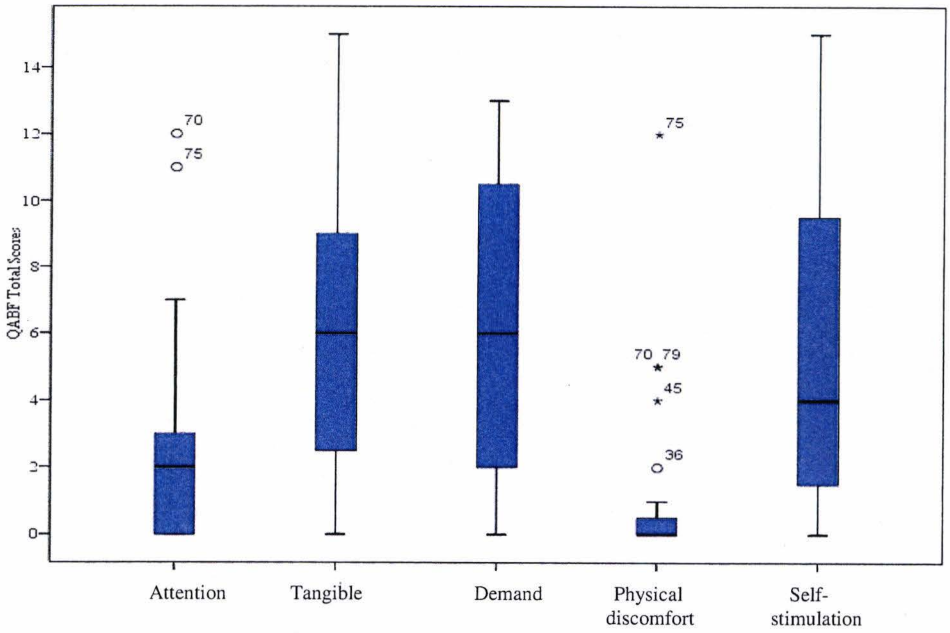
A series of Wilcoxon matched-pairs signed rank tests were conducted to compare pair-wise within group differences using a significance level of p = .005 (Bonferroni adjustment). The results of each pair-wise comparison are presented in Table 4.7 below. For aggressive behaviours the following pair-wise differences were significant at the .005 level: attention<tangible; attention<demand; attention>self-stimulatory; tangible>physical discomfort; tangible>self-stimulatory; demand>physical discomfort; demand>self-stimulatory.

Table 4.7

Results of Pair-wise Comparisons on the QABF for Children with FXS who Display Aggression

Pair-wise comparison	Wilcoxon's W	N (excluding ties)	*p* (*sig at .005 level)
Attention vs. Tangible	33	31	.000*
Attention vs. Demand	27.5	32	.000*
Attention vs. Physical discomfort	176.5	28	.555
Attention vs. Self-stimulation	60	25	.004*
Tangible vs. Demand	72	26	.007
Tangible vs. Physical discomfort	37.5	30	.000*
Tangible vs. Self-stimulation	13.5	29	.000*
Demand vs. Physical discomfort	.00	29	.000*
Demand vs. Self-stimulation	.00	29	.000*
Physical discomfort vs. Self-stimulation	69	22	.062

Figure 4.3. Boxplot for QABF total scores. Destructive behaviours



For destructive behaviours, the Friedman test was significant at the .016 level: χ^2 (4) = 28.325; p = 0.000.

A series of Wilcoxon matched-pairs signed rank tests were conducted to compare pair-wise within group differences using a significance level of p = .005 (Bonferroni adjustment). The results of each pair-wise comparison are presented in Table 4.8 below. For destructive behaviours the following pair-wise differences were significant at the .005 level: tangible>physical discomfort; demand>physical discomfort; physical discomfort<self-stimulation.

Table 4.8

Results of Pair-wise Comparisons on the QABF for Children with FXS who Display Destructive Behaviour

Pair-wise comparison	Wilcoxon's W	N (excluding ties)	*p* (*sig at .005 level)
Attention vs. Tangible	26	18	.007
Attention vs. Demand	42	20	.016
Attention vs. Physical discomfort	18.5	14	.029
Attention vs. Self-stimulation	59.5	20	.090
Tangible vs. Demand	37	12	.892
Tangible vs. Physical discomfort	11.5	20	.000*
Tangible vs. Self-stimulation	133.0	23	.444
Demand vs. Physical discomfort	9.5	19	.000*
Demand vs. Self-stimulation	129	23	.794
Physical discomfort vs. Self-stimulation	25.5	19	.003*

SMS. Within-group analysis.

Table 4.9 and Figures 4.4-4.6 depict the within-syndrome profile of scores on the QABF for children with SMS across self-injurious behaviour, aggression and destructive behaviours. There appears to be little differentiation between the five subscales for self-injurious behaviours, and with the exception of the self-stimulation subscale, for aggressive behaviours. It appears that there may be elevated scores for the attention subscale of the QABF for destructive behaviours, in comparison to other subscales.

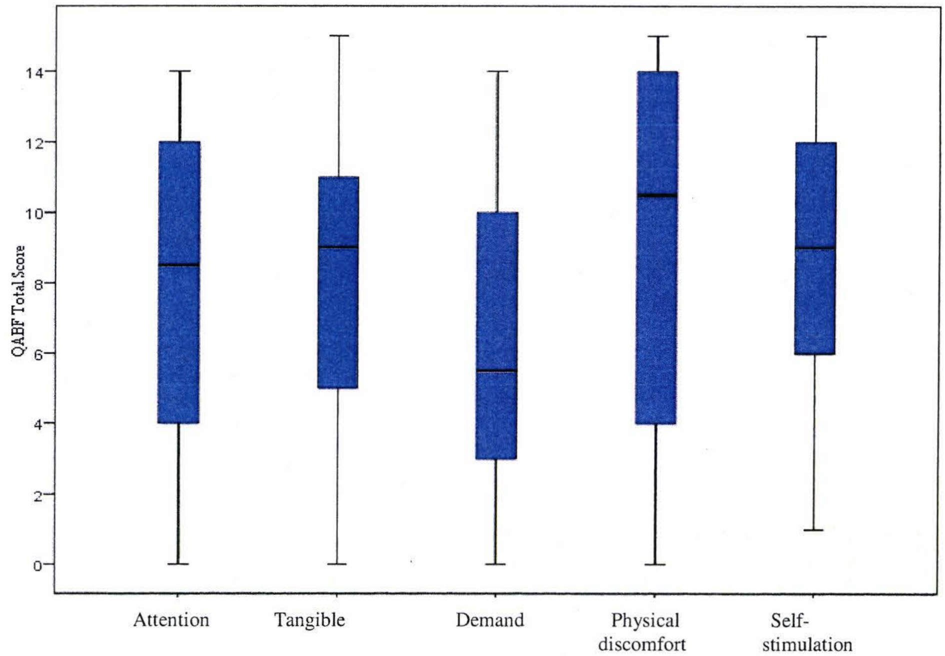
The Kolmogorov-Smirnov test of goodness-of-fit was not significant for any topography or subscale, suggesting the data were normally distributed. However, Mauchly's test of sphericity was significant for destructive behaviours. As such a series of non-parametric Friedman tests were used to examine within group differences in behavioural function for all behavioural topographies, using a significance level of .016 (Bonferroni adjustment).

Table 4.9

Within-Syndrome Results of the QABF for Children with SMS (Total Scores).

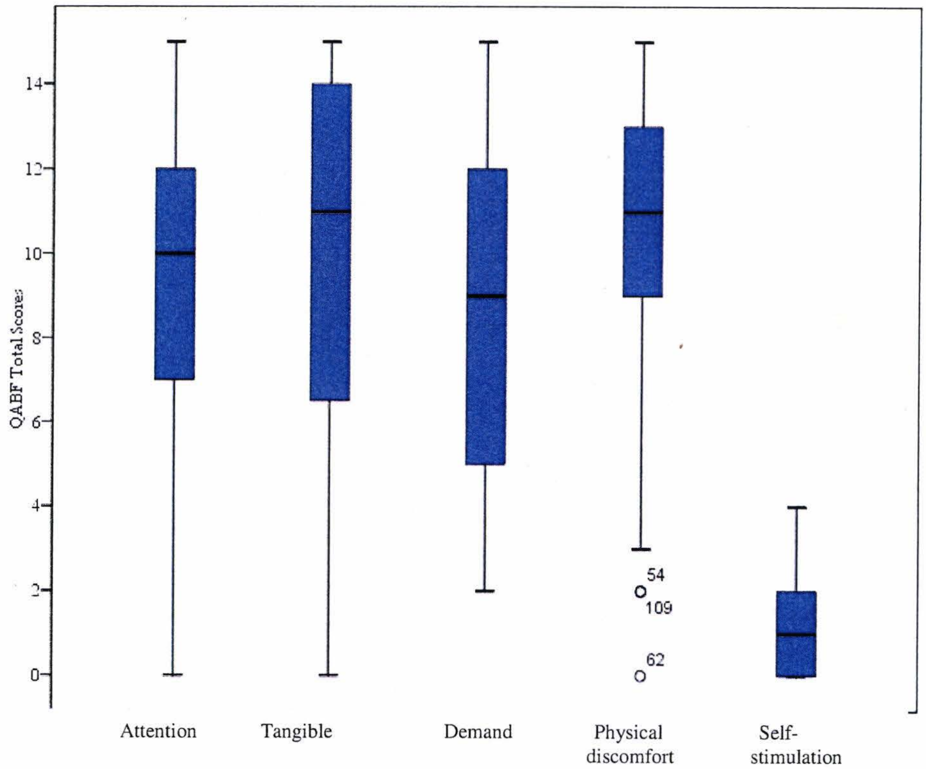
QABF Total Scores	Self-injurious behaviour (N=22)		Aggression (N=23)		Property Destruction (N=19)	
	M	SD	M	SD	M	SD
Attention Subscale	7.91	4.68	8.83	4.35	10.42	3.58
Tangible Subscale	8.23	4.47	9.87	4.77	9.05	4.18
Demand Subscale	6.36	4.19	8.39	4.38	8.26	4.45
Physical discomfort Subscale	9.05	5.33	10.00	4.61	9.26	5.42
Automatic Subscale	8.45	3.93	1.17	1.40	6.89	5.18

Figure 4.4. Boxplot for QABF total scores. Self-injurious behaviours



For self-injurious behaviours, the Friedman test was not significant at the .016 level: $\chi^2(4) = 3.529$; $p = 0.473$. Therefore, the null hypothesis that there were no within-group differences could not be rejected.

Figure 4.5. Boxplot for QABF total scores. Aggression



For aggressive behaviours, the Friedman test was significant at the .016 level: $\chi^2(4) = 40.724$; $p = 0.000$.

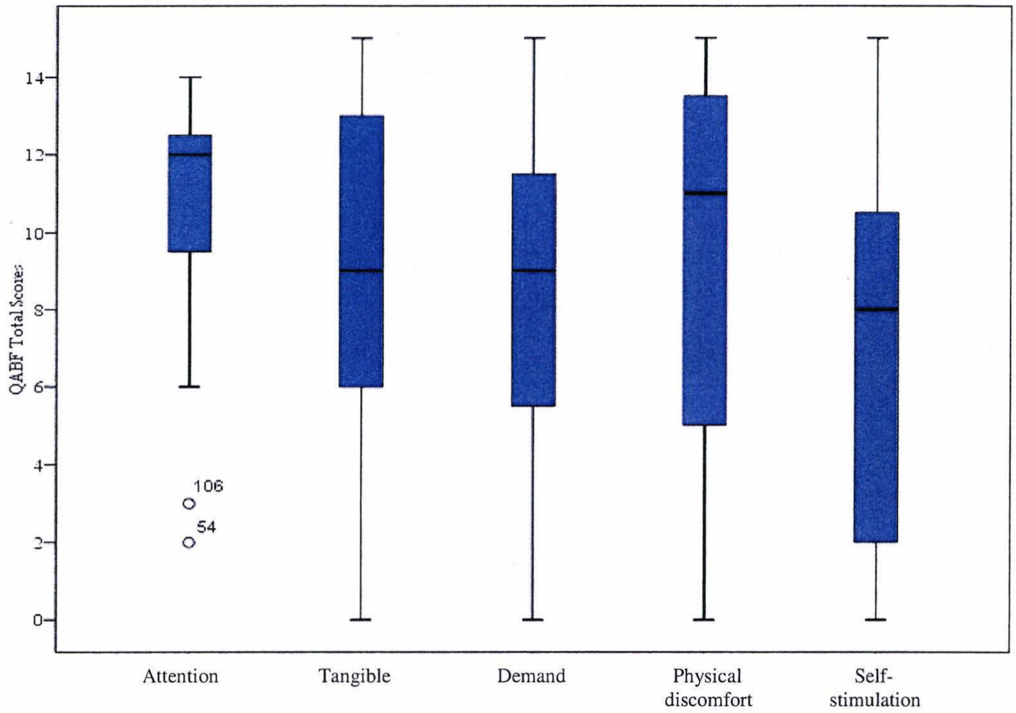
A series of Wilcoxon matched-pairs signed rank tests were conducted to compare pair-wise within group differences using a significance level of $p = .005$ (Bonferroni adjustment). The results of each pair-wise comparison are presented in Table 4.10 below. For aggressive behaviours the following pair-wise differences were significant at the .005 level: attention>self-stimulatory; tangible>self-stimulatory; demand>self-stimulatory; physical discomfort>self-stimulatory.

Table 4.10

Results of Pair-wise Comparisons on the QABF for Children with SMS who Display Aggressive Behaviour

Pair-wise comparison	Wilcoxon's W	N (excluding ties)	*p* (*sig at .005 level)
Attention vs. Tangible	82.0	20	.389
Attention vs. Demand	98.5	21	.553
Attention vs. Physical discomfort	75.5	20	.269
Attention vs. Self-stimulation	4	22	.000*
Tangible vs. Demand	79.5	21	.207
Tangible vs. Physical discomfort	111	21	.875
Tangible vs. Self-stimulation	0	21	.000*
Demand vs. Physical discomfort	91	22	.248
Demand vs. Self-stimulation	0	23	.000*
Physical discomfort vs. Self-stimulation	5	23	.000*

Figure 4.6. Boxplot for QABF total scores. Destructive behaviours.



For destructive behaviours, the Friedman test was not significant at the .016 level: $\chi^2(4) = 6.689$; $p = 0.153$. Therefore, the null hypothesis that there were no within-group differences could not be rejected.

Questions About Behavioral Function. Between-group analysis

For self-injurious behaviours there were a total of 30 participants with FXS, 22 with SMS and 24 from the mixed etiology group. For aggressive behaviours there were a total of 32 participants from the FXS group, 23 from the SMS group and 28 from the mixed etiology group. Finally for destructive behaviours there were a total of 23 participants from the FXS group, 19 from the SMS group and 15 from the mixed etiology group.

The Kolmogorov-Smirnov test of goodness-of-fit was significant at the .05 level for both FXS and the mixed etiology group across the physical discomfort and self-stimulatory subscales, suggesting that the data were not normally distributed on these subscales. As such a series of non-parametric Kruskal-Wallis tests were used to examine group differences across each subscale, using a significance level of .003 (Bonferroni adjustment).

Attention subscale.

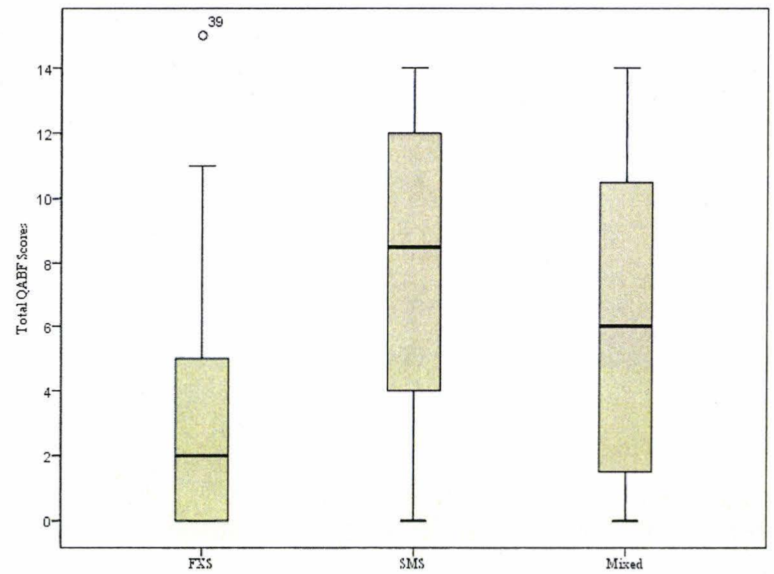
Table 4.11 and Figures 4.7-4.9 depict the between group comparison on the attention subscale of the QABF for self-injurious behaviour, aggression and property destruction. The groups appear to differ in a similar way across all topographies, with children with FXS consistently scoring lower than both comparison groups and children with SMS consistently scoring higher than either comparison group.

Table 4.11

Between-group Comparison of the QABF. Attention Subscale.

Attention subscale	FXS		SMS		Mixed Etiology	
	M	SD	M	SD	M	SD
Self-injurious behaviour	3.20	3.77	7.91	4.68	6.38	4.93
Aggression	3.62	3.93	8.83	4.35	6.61	4.88
Property destruction	2.78	3.46	10.42	3.58	6.33	4.60

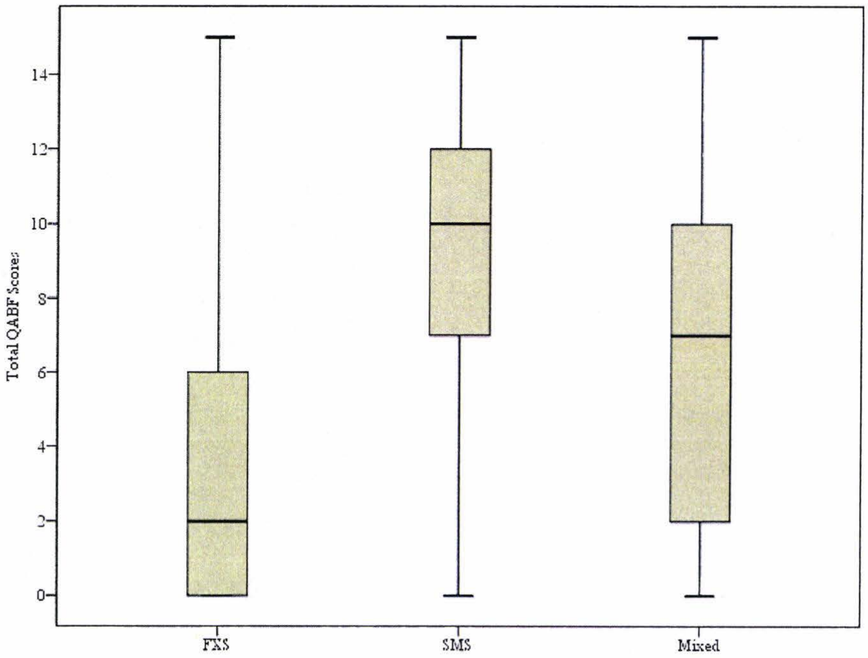
Figure 4.7. Boxplot for between-group QABF total scores for the attention subscale. Self-injury



For self-injury, the Kruskal-Wallis revealed significant between group differences on the Attention subscale at the .003 level: χ^2 (2) = 13.615; p = 0.001. Using a significance level of .016 (Bonferroni adjustment) a series of Mann-Whitney tests showed: differences between children with FXS and SMS were significant (U = 138.0; p = .000). Differences between children with FXS and the mixed etiology group were significant (U= 222.5; p = 0.015). Differences between the SMS group and mixed etiology group were not significant (U= 216.5; p = .294).

In sum, the following differences were significant for self-injury on the attention subscale of the QABF: FXS<SMS; FXS<mixed etiology.

Figure 4.8. Boxplot for between-group QABF total scores for the attention subscale. Aggressive behaviours.

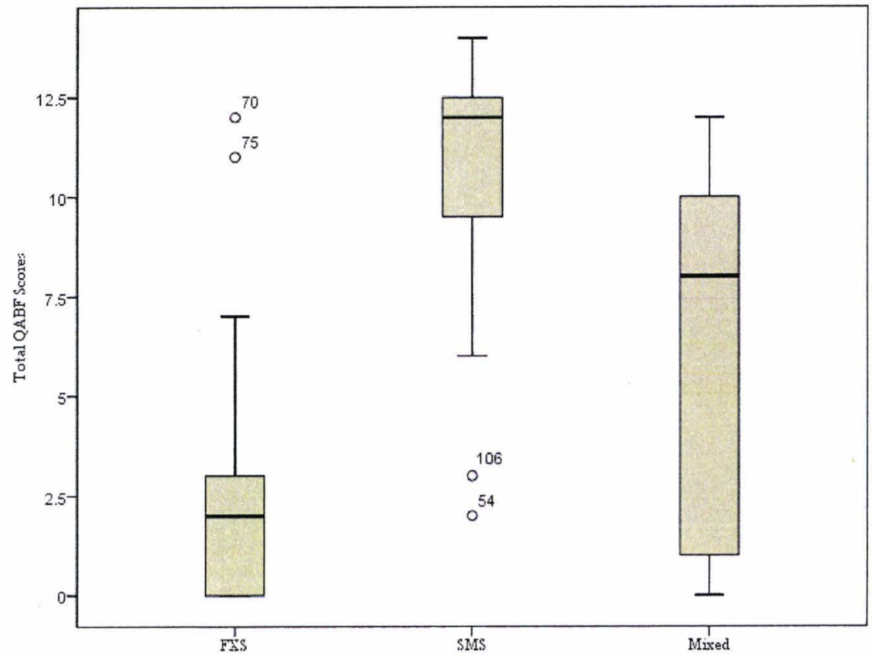


For aggression, the Kruskal-Wallis revealed significant between group differences on the Attention subscale at the .003 level: χ^2 (2) = 16.434; p = 0.000. Using a significance level of .016 (Bonferroni adjustment) a series of Mann-Whitney tests showed: differences between children with FXS and SMS were significant (U = 137.5; exact p = .000), as were differences between children with FXS and the mixed etiology group (U= 285.0; p = 0.015). Differences between the SMS group and mixed etiology group were not significant (U= 234.5; exact p = .096).

In sum, the following differences were significant for aggression on the attention subscale of the QABF: FXS<SMS; FXS<mixed etiology.

Figure 4.9. Boxplot for between-group QABF total scores for the attention subscale.

Destructive behaviours



For destructive behaviour, the Kruskal-Wallis revealed significant between group differences on the Attention subscale at the .003 level: $\chi^2(2) = 23.553$; p = 0.00. Using a significance level of .016 (Bonferroni adjustment) a series of Mann-Whitney tests showed: differences between children with FXS and SMS were significant (U = 36.5; p = .000), as were differences between the SMS group and mixed etiology group (U= 58.5; p = .003). Differences between children with FXS and the mixed etiology group were non-significant (U = 108.0; p =.050).

In sum, the following differences were significant for destructive behaviours on the attention subscale of the QABF: FXS<SMS; SMS>mixed etiology.

Tangible subscale.

Table 4.12 depicts the between group comparison on the tangible subscale of the QABF for self-injurious behaviour, aggression and property destruction. The groups appear to have a relatively similar profile of scores across topographies.

Table 4.12

Between-group Comparison of the QABF. Tangible Subscale

Tangible subscale	FXS		SMS		Mixed Etiology	
	M	SD	M	SD	M	SD
Self-injurious behaviour	8.57	3.74	8.23	4.47	7.29	4.69
Aggression	8.16	4.09	9.87	4.77	10.25	4.21
Property destruction	5.87	4.29	9.05	4.18	10.00	3.96

For self-injurious behaviour, the Kruskal-Wallis revealed no significant between group differences on the tangible subscale at the .003 level: χ^2 (2) = 1.031; p = .597.

For aggressive behaviour there were no significant between group differences on the tangible subscale at the .003 level: χ^2 (2) = 4.587; p = .101.

For destructive behaviour, there were no significant between-group differences on the tangible subscale at the .003 level: χ^2 (2) = 8.915; p = .012.

Demand.

Table 4.13 depicts the between group comparison on the demand subscale of the QABF for self-injurious behaviour, aggression and property destruction. There appears to be few between-group differences on this subscale.

Table 4.13

Between-group Comparison of the QABF. Demand Subscale

Demand subscale	FXS		SMS		Mixed Etiology	
	M	SD	M	SD	M	SD
Self-injurious behaviour	9.07	3.79	6.36	4.19	7.71	5.50
Aggression	10.12	3.99	8.39	4.38	10.46	4.13
Property destruction	6.04	4.48	8.26	4.45	8.60	4.14

For self-injurious behaviours, the Kruskal-Wallis was not significant at the .003 level: χ^2 (2) = 4.065; p = .131.

For aggressive behaviours, the Kruskal-Wallis was not significant at the .003 level: χ^2 (2) = 3.656; p = .161.

For destructive behaviours there were no significant between group differences on the demand subscale at the .003 level: χ^2 (2) = 3.191; p = .203.

Physical discomfort.

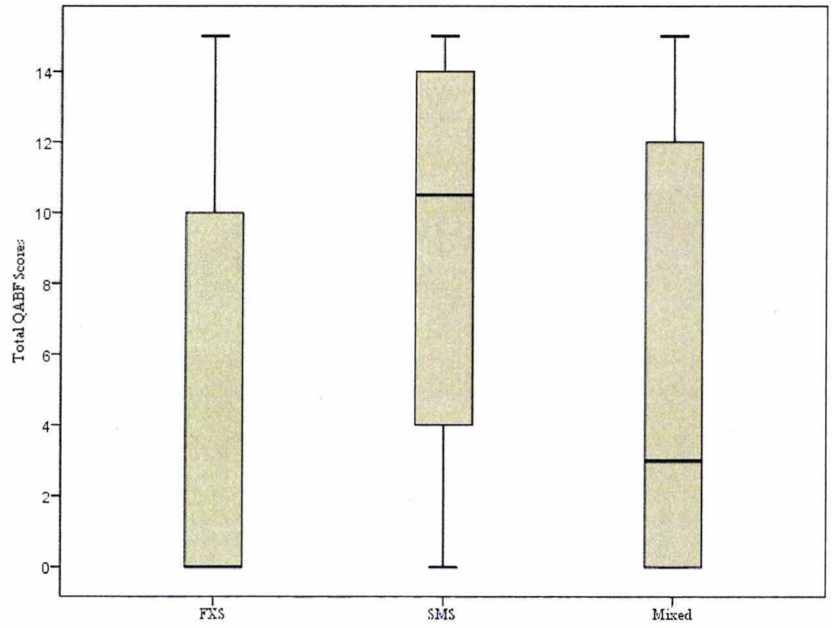
Table 4.14 and Figures 4.10-4.12 depict the between group comparison on the physical discomfort subscale of the QABF across each topography. The groups appear to differ in a similar way across all topographies, with children with FXS consistently scoring lower than both comparison groups and children with SMS consistently scoring higher than either comparison group.

Table 4.14

Between-group Comparison of the QABF. Physical Discomfort Subscale

Physical discomfort subscale	FXS		SMS		Mixed Etiology	
	M	SD	M	SD	M	SD
Self-injurious behaviour	3.87	5.37	9.05	5.33	5.21	5.93
Aggression	3.22	4.65	10.0	4.61	4.75	5.58
Property destruction	1.26	2.85	9.26	5.42	3.73	5.31

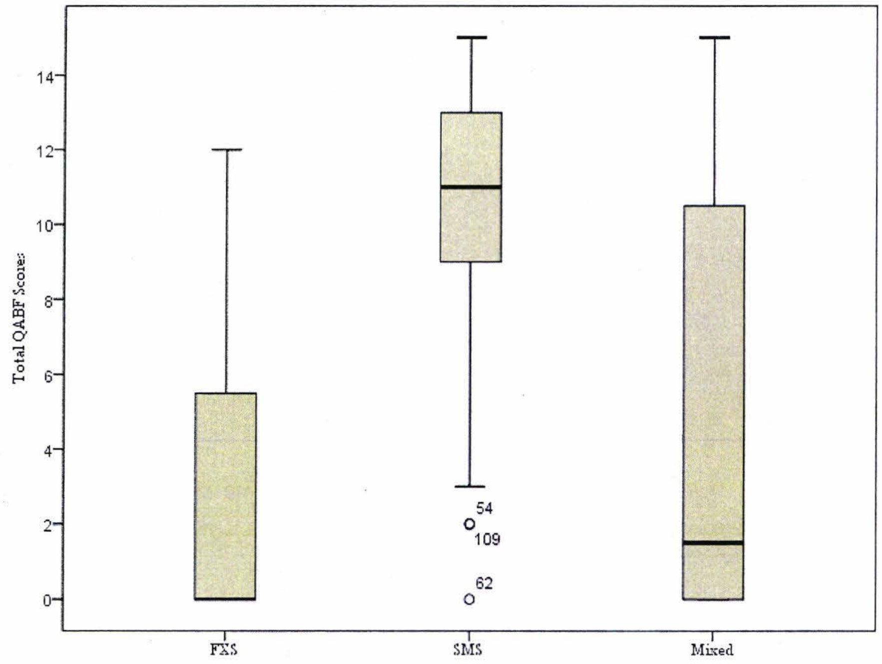
Figure 4.10. Boxplot for between-group QABF total scores for the physical discomfort subscale. Self-injurious behaviours



For self-injurious behaviour, the Kruskal-Wallis revealed significant between group differences on the physical discomfort subscale at the .003 level: χ^2 (2) = 12.482; p = 0.002. Using a significance level of .016 (Bonferroni adjustment) a series of Mann-Whitney tests showed: differences between children with FXS and SMS were significant (U = 154.0; p = .001). Differences between children with FXS and the mixed etiology group were non-significant (U= 275.5; p =.120). Differences between the SMS group and mixed etiology group were not significant (U= 162.0; p = .024).

In sum, the following differences were significant for self-injury on the physical discomfort subscale of the QABF: SMS>FXS.

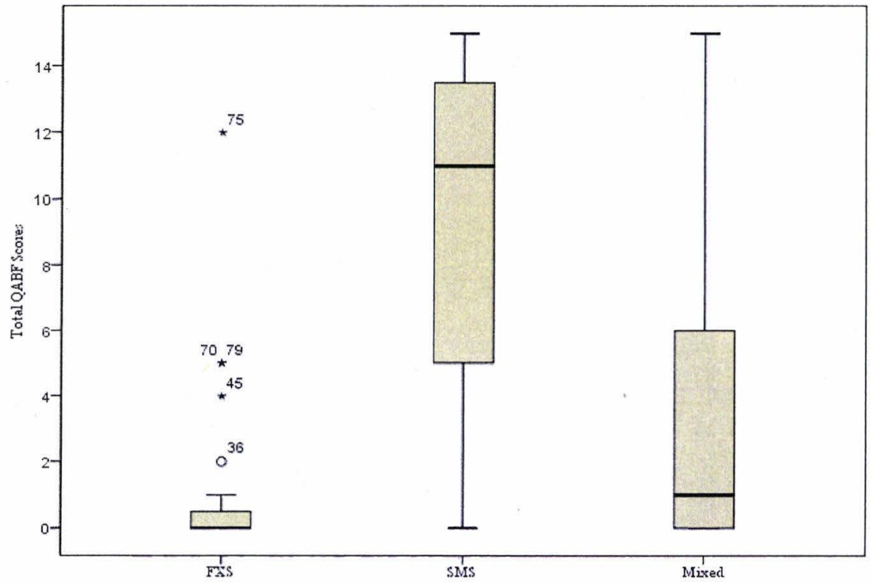
Figure 4.11. Boxplot for between-group QABF total scores for the physical discomfort subscale. Aggressive behaviours.



For aggressive behaviour, the Kruskal-Wallis revealed significant between group differences on the physical discomfort subscale at the .003 level: χ^2 (2) = 21.93; p = 0.000. Using a significance level of .016 (Bonferroni adjustment) a series of Mann-Whitney tests showed differences between children with FXS and SMS were significant (U = 114.5; p = .000), as were differences between the SMS group and mixed etiology group (U = 152.5; p = .001). Differences between children with FXS and the mixed etiology group were non-significant (U = 354.5; exact p =.143).

In sum, the following differences were significant for aggression on the physical discomfort subscale of the QABF: SMS>FXS; SMS>mixed etiology.

Figure 4.12. Boxplot for between-group QABF total scores for the physical discomfort subscale. Destructive behaviours.



For destructive behaviours, the Kruskal-Wallis revealed significant between group differences on the physical discomfort subscale at the .003 level: χ^2 (2) = 21.598; p = 0.000. Using a significance level of .016 (Bonferroni adjustment) a series of Mann-Whitney tests showed: differences between children with FXS and SMS were significant (U = 48.5; p = .000), as were differences between the SMS group and mixed etiology group (U = 66.5; p = .008). Differences between children with FXS and the mixed etiology group were non-significant (U = 120.0; p =.070).

In sum, the following differences were significant for destructive behaviours on the physical discomfort subscale of the QABF: SMS>FXS; SMS>mixed etiology.

Automatic.

Table 4.15 depicts the between group comparison on the automatic subscale of the QABF for self-injurious behaviour, aggression and property destruction. The groups appear to have a relatively similar profile of scores across topographies.

Table 4.15.

Between-group Comparison of the QABF. Automatic Subscale

Automatic subscale	FXS		SMS		Mixed Etiology	
	M	SD	M	SD	M	SD
Self-injurious behaviour	6.13	4.29	8.45	4.35	4.96	4.81
Aggression	1.23	2.23	1.17	1.40	1.64	2.66
Property destruction	5.65	5.31	6.89	5.18	2.93	3.35

For self-injurious behaviour, the Kruskal-Wallis was not significant at the .003 level: χ^2 (2) = 6.653; p = .036.

For aggressive behaviours, the Kruskal-Wallis was not significant at the .003 level: χ^2 (2) = .314; p = .855.

For destructive behaviours, the Kruskal-Wallis was not significant at the .003 level: χ^2 (2) = 4.832; p = .089.

Categorical Analysis of Between-group Differences in Behavioural Function.

An additional analysis was also conducted to examine whether the same differences on the QABF were also present when the data were analysed categorically. A function was deemed to be present only if the score on a subscale was 10 or more (requiring that a minimum of 4/5 of items on the QABF were endorsed for a specific subscale).

Between-group differences were examined for each of the five subscales across each topography. Using a significance level of 0.003 (Bonferroni adjustment) a series of chi square analyses were conducted. For self-injurious behaviours, there were no significant between-group differences on either scale, there was however a non-significant trend for the attention subscale (p = .010). For both aggression and property destruction significant differences were found on the attention and physical discomfort subscales of the QABF.

Table 4.16 and Table 4.17 show contingency tables for the attention and physical discomfort subscales of the QABF showing nominal data for the three groups across each topography. All percentages refer to differences within each diagnostic group.

Although there were some discrepancies between the two analyses, specifically on the findings for the self-injurious behaviour subscale, these categorical findings are broadly consistent with those reported in the previous section. A lower proportion of children with FXS were reported to display attention-maintained challenging behaviours. A higher proportion of children with SMS were reported to display challenging behaviours related to physical discomfort. This provides further evidence to suggest that the three groups differed solely on scores on the attention and physical discomfort subscales of the QABF.

Table 4.16.

Categorical Data for Attention Subscale of the QABF by Topography

Topography	FXS	SMS	Mixed Etiology
Attention subscale	N (%)	N (%)	N (%)
Self-injury			
Yes	2 (6.7%)	9 (40.9%)	8 (33.3%)
No	28 (93.3%)	13 (59.1%)	16 (66.7%)
Aggression			
Yes	2 (6.2%)	14 (60.9%)	9 (32.1%)
No	30 (93.8%)	9 (39.1%)	19 (67.9%)
Destructive			
Yes	2 (8.7%)	14 (73.7%)	6 (40%)
No	21 (91.3%)	5 (26.3%)	9 (60%)

Table 4.17

Categorical Data for the Physical Discomfort Subscale of the QABF by Topography

Topography/Subscale	FXS	SMS	Mixed Etiology	
Physical Discomfort subscale		N (%)	N (%)	N (%)
Self-injury				
Yes	9 (30%)	12 (54.5%)	7 (29.2%)	
No	21 (70%)	10 (45.5%)	17 (70.8%)	
Aggression				
Yes	7 (21.9%)	17 (73.9%)	8 (28.6%)	
No	25 (78.1%)	6 (26.1%)	20 (71.4%)	
Destructive				
Yes	1 (4.3%)	12 (63.2%)	3 (20%)	
No	22 (95.7%)	7 (36.8%)	12 (80%)	

Summary of Results

Behavioural Form

- The SMS group had significantly higher scores on the Communication sub-domain of the Vineland Screener than either comparison group.
- There were no other between group differences on Vineland Screener scores.
- There were no significant differences between the groups on any subscale of the ABC-C. There was a non-significant trend on the Irritability subscale, with children with FXS appearing to have lower scores than the SMS group.
- The SMS group showed significantly higher scores on the BPI SIB subscale than did the FXS group.

Behavioural Function

Within group.

- The FXS group showed significantly lower scores on the attention subscale than tangible or demand for both self-injurious behaviour and aggressive behaviours, there was a non-significant trend in the same direction for destructive behaviours.
- The FXS group showed significantly lower scores on the physical discomfort subscale than tangible or demand for self-injurious behaviour, aggression and destructive behaviours.
- Both the FXS and SMS groups showed significantly lower scores on the automatic subscale for aggressive behaviours than all other social subscales.
- There were no other within-group differences for the SMS group.

Between group.

- The FXS group had significantly lower scores on the attention-subscale than the SMS group for all three topographies.
- The FXS group had significantly lower scores on the attention-subscale than the mixed etiology group for self-injurious and aggressive behaviours. There was a non-significant trend in the same direction for destructive behaviours.
- The SMS group had significantly higher scores than the mixed etiology group on the attention subscale for destructive behaviour.
- The SMS group had significantly higher scores on the physical discomfort subscale of the QABF than the FXS group or mixed etiology group for aggressive and destructive behaviours, and only the FXS group for self-injurious behaviour.
- There were no between group differences for the tangible, demand or automatic subscales across any topographies.

- Categorical analyses were broadly supportive of these findings. Significant differences were found between the groups only on the attention and physical discomfort subscales of the QABF for aggression and property destruction.

Discussion

The current study found within- and between-group differences in both the form and function of adaptive and challenging behaviours in children with FXS and SMS. Using the QABF as a measure of behavioural function the results of both the within- and between-group comparison suggested that children with FXS were less likely to display attention-maintained challenging behaviours across all topographies. The results of the between-group comparison showed that children with SMS were more likely to display challenging behaviours related to physical discomfort across all topographies than were other groups. To the author's knowledge these are the first reported between-syndrome differences in the function served by challenging behaviour.

The results of the current study suggested that the groups were relatively well matched for CA and, with the communication sub-domain of the Vineland Screener apart, adaptive behaviour. Prior studies have suggested that children with SMS tend to score lower on the Daily Living Skills sub domain than would typically be expected given levels of functioning in other areas (Greenberg et al., 1996; Madduri et al., 2006; Udwin, Webber, & Horn, 2001); although some studies have failed to replicate this finding (Dykens, Finucane, & Gayley, 1997). Daily living skills have also been reported to be a relative area of strength for people with FXS in comparison to communication and social skills (Dykens et al., 1989; Dykens, Ort et al., 1996; Hatton et al., 2003); although again some studies have failed to find such differences (Baumgardner, Reiss, Freund, & Abrams, 1995). The current study showed that children with SMS were reported to have a higher level of communication skills than either control group. There were no significant differences for either children with SMS or children with FXS on scores for any other sub-domain of the Vineland Screener. This is the first study to compare adaptive behaviour in FXS and SMS against one another and against a mixed etiology control.

There were no significant differences between the groups in carer reports of the severity of challenging behaviour. There was a non-significant trend however for the Irritability subscale of the ABC-C, with children with SMS appearing to score higher than the FXS group. Relatively high scores on the Irritability subscale of the ABC in comparison to other groups have been reported in prior research involving people with SMS (Clarke & Boer, 1998). Other studies of challenging behaviour in SMS suggest relatively high levels of similar problems. For example Dykens et al (1997) reported high scores on the Aggressive Behaviour domain of the Reiss Screen in their sample of people with SMS. Previous studies to have used the ABC with males with FXS have reported significantly higher scores on the Hyperactivity, Inappropriate Speech and Stereotypical Behaviour subdomains in comparison to mixed etiology controls (Baumgardner et al., 1995). No such differences were found for the FXS group in the current study, however.

Using the BPI- SIB subscale, children with SMS were reported to display more frequent self-injurious behaviour than children with FXS. Although comparisons with the results of other prevalence studies have suggested higher levels of self-injury in people with SMS than would be typically expected (Sloneem, 2005), to the author's knowledge there have been no other direct between group comparisons of self-injurious behaviour involving children with SMS.

Both within- and between-syndrome differences were found in the function served by challenging behaviour. These are of particular interest as the current study is the first to have included a between groups comparison of behavioural function in either of these syndrome groups. The only subscales on which significant between-group differences were found were the attention and physical discomfort subscales of the QABF.

Children with FXS appeared to be less likely to display challenging behaviours that were reported to be maintained by social attention than children with SMS or the mixed etiology group. This conclusion was supported by the within-syndrome profile of scores for the QABF across each topography. There was also some indication in the within-group analysis that children with FXS were more likely to display aggressive behaviours that were maintained by escape than any other function. In short, children with FXS appeared to be less likely to display attention maintained challenging behaviour, than other functions (particularly escape-maintained). This profile reflects existing findings that have been reported in the literature. Using a measure derived from the Functional Assessment Interview (O'Neil, Horner, Albin, Storey, & Sprague, 1990), Symons (2003), reported that only 3% of boys with FXS who displayed self-injurious behaviour were reported to do so in order to access attention, in comparison to 87% of participants who did so in response to routine changes, and 65% in response to task demands. Other studies have demonstrated that children with FXS may be particularly likely to engage in challenging behaviours in situations that are characterised by high social or performance related demands (Hall et al., 2006; Hessl, Glaser, Dyer-Friedman, & Reiss, 2006; Lesniak-Karpiak, Mazzocco, & Ross, 2003; Woodcock et al., 2009).

It is unclear as to why children with FXS may be less likely to display attention-maintained challenging behaviours. One mechanism that has been postulated is that FXS is associated with the abnormal functioning of the limbic-hypothalamic-pituitary-adrenal (LHPA) axis, which plays an important role in the human stress response (Hessl et al., 2002). Further work is needed to examine the influence of such physiological variables on the function of challenging behaviour.

There was evidence to suggest that children with SMS were more likely to display attention-maintained challenging behaviours than children with FXS and attention-maintained destructive behaviours than mixed-etiology controls.

Previous studies have highlighted an apparent heightened 'need' for adult attention in individuals with SMS (Dykens et al., 1997; Smith et al., 1998). A within-syndrome study using the QABF reported that individuals with SMS were particularly likely to display attention-maintained physical and verbal aggression (Sloneem, 2005). Likewise, in a recent study Taylor and Oliver (2008) reported that the challenging behaviour of four of five children with SMS was more likely to occur following periods of low adult attention or following reduced levels of demands. Challenging behaviour was, for these individuals, likely to lead to an increase in attention or demands. Taylor and Oliver suggest that children with SMS may have a "predisposition to experience social or other stimuli...as significantly rewarding" (p. 839). The current study, although hampered by low N, provides some preliminary evidence to support this assertion. The within-group analysis for children with SMS did not however, show any clear differentiation between the different subscales suggesting that children with SMS may be likely to display challenging behaviour that is multiply controlled.

The current study found that in comparison to other groups, children with SMS were more likely to display challenging behaviour related to physical discomfort. Prior studies have suggested a possible relationship between health factors in SMS, such as chronic sleep deprivation, and challenging behaviour (De Leersnyder et al., 2001). However the current study is the first to identify between group differences in such relations using a measure of behavioural function. Given the high occurrence of sleep disturbance and peripheral neuropathy in SMS (Finucane et al., 2001; Greenberg et al., 1996), then high levels of challenging behaviour related to physical discomfort should perhaps be expected in this group. Whilst further experimental work is required to delineate the nature of such interactions, treatment of such conditions would also represent an obvious first step for clinicians working with individuals with SMS.

There are a number of limitations with the current study which may hamper the strength of conclusions which can be drawn from the findings presented above.

The measure of behavioural function was indirect and it may be that the responses of parents/caregivers do not correspond to the actual contingencies that influence the behaviour of their child. Further research is required which adopts experimental functional analysis methods in order to provide a more rigorous examination of behavioural function than was possible in the current study.

It is unclear whether social attention functioned as a less effective type of reinforcement for children with FXS (i.e., its value as a type of reinforcement is enduringly abolished), or whether it functioned as an aversive stimulus (i.e., its value as a type of punishment is enduringly established). The use of an alternative measure of behavioural function which includes a social-escape subscale may have helped tease such information out. This hypothesis could be explored using experimental analogues. For example, high levels of challenging behaviour in a condition characterised by high levels of attention, with escape provided contingent on challenging behaviour, would indicate that social attention functioned as an aversive stimulus. In contrast, low levels of challenging behaviour in this same condition and in the standard attention condition, would indicate that rather than functioning as an aversive stimulus, attention was simply an ineffective source of reinforcement.

There were a number of problems with participant recruitment which may have limited the power of the statistical analysis involving the SMS group. In addition, the reliance on convenience sampling (primarily via parental support groups) may have impinged on the external validity of these findings. For example, it is possible that parents of a child who displayed more severe challenging behaviour were more likely to volunteer to participate in the study. Whilst potentially limiting the generalisability of the current findings, there is no data of which the author is aware to suggest that topographical severity is related to behavioural function, suggesting that this may have limited implications for the interpretation of findings relating to the QABF.

Previous research suggests that females with FXS may be less affected than males and differ in terms of the severity of challenging behaviour (Hessl et al., 2002). Other studies of challenging behaviour in this syndrome have tended to treat males and females separately for this reason. However, due to the emphasis of the current study on behavioural function and in order to facilitate comparisons with the SMS group, females with FXS were included in the current study. Only three females with FXS took part in the current study, precluding the use of inferential statistics to examine any potential gender differences in behavioural function. However, inspection of descriptive statistics did not suggest any notable gender differences in scores on the QABF subscales for children with FXS.

The inclusion of several different topographies within each general category of behaviour, may have led to behaviours that formed separate response classes being treated together. It is recommended that the QABF be completed for each separate topography of challenging behaviour in order to prevent such problems (Matson & Vollmer, 1995). Whilst this would have allowed for a more sensitive analysis, the differential pattern of results across each behavioural function suggests that the methods adopted in the current study were still sufficient to determine general differences in the probability of certain functions being endorsed.

These weaknesses aside, the findings of the current study are promising and should stimulate further research on the influence of genetic events on behavioural function. Experimental analogue methods would appear to be a logical next step in order to overcome some of the limitations identified in the current study. Future research is also required that examines the influence of third variables (or the 'endophenotype') on behaviour-environment relations. For example, it would be of interest to examine the extent to which variations in the functioning L-HPA axis underpin the behaviour-environment relations identified above for children with FXS. Further study of the association between health-related factors associated with SMS and challenging behaviour would also seem to be required. Experimental analogue methods may also help in this regard and have been demonstrated to be especially effective at identifying the influence of factors such as sleep deprivation (cf., Kennedy & Meyer, 1996; O'Reilly, 1995).

The current study has provided some preliminary evidence of the role of GxE in challenging behaviour displayed by children with FXS and SMS. Evidence was provided to suggest that developmental changes associated with certain genetic syndromes may alter the reinforcing value of certain environmental consequences. Specifically, children with FXS were less likely to display attention-maintained challenging behaviour and children with SMS were more likely to display physical discomfort-related and possibly attention-maintained behaviour than would be typically expected. Such relations are likely to be particularly important to the development of challenging behaviour in children with certain genetic syndromes (Langthorne & McGill, 2008).

Chapter V.

Gene-Environment Interactions. An Experimental Analysis.

"The genesis of many events that function as positive or negative reinforcers has a phylogenetic basis and is expressed as behavioral phenotypes with unique topographies of behavior-environment patterns."(Kennedy, Caruso, & Thompson, 2001, p. 544)

Chapter Overview

It has been suggested that genetic events may influence the function served by challenging behaviour. The study reported in the previous chapter adopted indirect questionnaire methods to examine this question in children with FXS and SMS; reporting both within- and between-group differences in the function served by challenging behaviour. The study reported in the current chapter adopted experimental single-case design methodology to examine this same question. These methods were used to overcome some of the weaknesses associated with the use of indirect methods of functional assessment.

A subset of children from the FXS and SMS groups were included in the current study. Variations in the occurrence of challenging behaviour across different environmental conditions were examined for each child. This allowed for a comparison of the function served by challenging behaviour both within- and between- each group. Of the eight children with FXS; four were shown to display at least one response class of challenging behaviour that was tangible-maintained, five displayed at least one response class that appeared to be negatively-reinforced by the removal of demands or social attention, none displayed challenging behaviours that were found to be positively reinforced by the provision of social attention. Of the six children with SMS, four were found to display multiply controlled challenging behaviour which was, at least in part, attention-maintained. The implications of these findings for future research, as well as for the assessment and treatment of challenging behaviour are discussed.

Introduction

It has been suggested that, for some individuals, genetic events may enduringly alter the reinforcing value of the consequences that maintain challenging behaviour (Kennedy et al., 2001; Langthorne & McGill, 2008; Oliver, 1993). Evidence from a number of recent studies suggests that the challenging behaviour of individuals with FXS may be less likely to be maintained by social attention and more likely to be maintained by the removal of aversive stimuli, and/or the provision of tangibles (Hall, DeBernadis, & Reiss, 2006; Symons, Clark, Hatton, Skinner, & Bailey, 2003; Woodcock, Oliver, & Humphreys, 2009). In contrast, the challenging behaviour of children with SMS may be more likely to be attention-maintained (Dykens & Smith, 1998; Sloneem, 2005; A. Smith, Dykens, & Greenberg, 1998; Taylor & Oliver, 2008).

The study reported in the previous chapter reported between-syndrome differences in behavioural function between children with FXS and SMS. Specifically, the study showed that children with FXS were less likely to display attention-maintained challenging behaviour than children with SMS or controls. In addition, children with SMS were more likely to display pain-related behaviours than children with FXS or mixed etiology controls. The study also reported within-syndrome differences. Children with FXS were less likely to display attention-maintained challenging behaviours than escape- or tangible-maintained challenging behaviours. Interestingly, and contrary to findings reported in prior research (e.g., Taylor & Oliver, 2008), children with SMS were no more likely to display attention-maintained behaviours than either escape- or tangible-maintained behaviour.

There were a number of weaknesses associated with the above study. The study adopted an indirect measure of behavioural function. Whilst this enabled a comparison involving relatively large numbers of participants, care-giver perceptions of the determinants of challenging behaviour may not necessarily correspond to the actual contingencies responsible for the evocation and maintenance of challenging behaviour. As such, a comparison based on direct observation and experimental manipulation may provide a more rigorous examination of the research question. Secondly, it was unclear from the previous study as to whether attention functioned as an aversive stimulus for children with FXS (i.e., the value of attention is enduringly established as a type of punishment) or whether attention simply does not function as a particularly effective reinforcer for this group of children (i.e., the value of attention as a type of reinforcement is enduringly abolished). Analogue methods may allow further exploration of this by examining the occurrence of challenging behaviour in children with FXS when in situations characterised by high levels of social contact.

The methods used in study two differ markedly from those in study one and are rooted in a very different underlying epistemology[23]. Some time will therefore be spent describing the underpinnings of experimental single-case design and contrasting this against the group-comparison approach used in the study reported in the previous chapter. The use of experimental functional analysis methodology shall then be justified.

Experimental Single Case Design

Morgan and Morgan (2001) present a number of characteristics of single-case design which are used to structure the following discussion.

Repeated measurement of behaviour over time.

Single-case design, with few exceptions, relies upon the direct observation of changes in behaviour rather than indirect measures such as those used in study one. Measurement is 'direct' when the behaviour being measured is exactly the same as that which is of interest. This correspondence between the phenomena of interest and what is being measured is one of the main advantages of the methods used in the current study.

[23] Such differences have: "*to do with what counts as the subject matter of psychology, with what questions we should ask about this subject matter, with how we should go about finding answers to these questions, with the status of existing psychological knowledge, and with whether psychology can be a science.*" (Lee, 1992).

Rather than being a static entity, behaviour is continuous and changes over time. Group design methods such as those used in study one typically rely on single measures of dependent variables in relative temporal isolation and thereby fail to account for the complexity of their subject matter. In contrast single-case design requires the experimenter to remain in close contact with the ongoing process of behaviour[24].

The strength of the assertions that can be made about the effects of an independent variable on a dependent variable is directly related to the number of data points for which behaviour is observed. This contrasts markedly with the approach common in group designs of taking a small number of data points for a large number of participants and using a group 'average' to summarise group performance.

Participants serving as their own controls.

Of interest to the behavioural scientist are the effects of a particular experimental manipulation on the behaviour of a single participant. In this sense single case research is characterised by participants serving as their own controls. In contrast, control in study one was achieved by comparing the performance of an experimental and a control group. However as Johnston and Pennypacker (1993) note:

> *There is no such phenomenon as "group behaviour" because there is no such organism as the "group" or "average subject." If an effect appears in group but not in the individual records then, it should not be considered solely a result of experimental variables (p.304).*

This emphasis on a 'thorough analysis' of the behaviour of the individual represents the hallmark of single-case design. A stable pattern of behaviour prior to any experimental manipulation is termed a *steady state* and it is this that provides the baseline

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[24] As Sidman (1960) states: "*Behavioural processes occur in time and must be measured over time. To identify the precise boundaries of a process frequent measurements are necessary.*" (p. 288).

of control against which subsequent predictions and verifications can be made[25]. The baseline or control condition allows predictions to be made about the effect of an independent variable and this prediction can then be verified by an actual change in performance that occurs when the independent variable is introduced.

Emphasis on experimental replication.

Prediction and verification are necessary but not sufficient conditions for the demonstration of a functional relationship. Rather the replication of such effects both within and between individuals is a fundamental requirement of single-case design. Intra and inter-participant replication contributes to both the internal and external validity of single-case design. There are a host of potential threats to the internal validity of any pre-post test experiment; and it is through the process of systematic replication that a functional relationship can be demonstrated between an experimental manipulation and behavioural change. Group design methods, such as those used in study one rarely adopt experimental replication as a means of ensuring internal validity.

The external validity of group design research is typically judged on the representativeness of the sample and the extent to which it reflects the underlying population. This contrasts markedly to the demonstration of external validity in single-case design, in which the aim is to verify initial findings in '*ever widening set of conditions*' (Sidman, 1960) by systematically replicating any effects across individuals, settings and behaviours.

[25] As Sidman (1960) states: *"The descriptive investigation of steady-state behavior must precede any manipulative study. Manipulation of new variables will often produce behavioural changes, but in order to describe the changes we must be able to specify the baseline from which they occurred; otherwise we face insoluble problems of control, measurement and generality.*" (p. 238).

Graphic presentation and visual analysis of data.

Group-design research typically involves using inferential statistics to determine whether observed between- or within-group differences are of a sufficient magnitude to reject the null hypothesis that no difference exists between the underlying population parameters. Johnston and Pennypacker (1993) suggest that the use of inferential statistics is ill-suited to the study of behavioural change. Whilst inferential statistics may be useful in supporting an analysis of behavioural processes they should not be used as the primary means of data analysis in single case design. Instead the existence of a functional relationship is best determined by visual inspection. Such an approach allows the experimenter to remain in close contact with the behaviour of his subject, and prevents potentially interesting data being lost within a myriad of statistical procedures.

Unique treatment of variability.

Radical behaviourism assumes human behaviour is lawful. As such rather than treating variability as 'experimental noise' to be silenced through the use of statistical procedures, it is assumed that variability simply reflects functional relationships that are yet to be uncovered (Sidman, 1960).

Unexplained sources of variability function as a barrier to establishing generality, and radical behaviourism therefore accepts the individuality of the organism rather than suppressing it in favour of the average (Chiesa, 1992). The aim of an experimental analysis is therefore to explain the sources of such variability and thereby transform such 'confounds' into independent variables in their own right.

In sum, the characteristics of experimental single-case design and the logic which underpins it stand in stark contrast to the group-comparison methods adopted in study one. Group comparison and statistical analysis may be of use as a supplemental aid to studying behavioural processes. However of primary importance is the demonstration of functional relationships at the level of the individual arrived at through an inductive process of prediction, verification and replication. Such experimental tactics are utilised in study two in the form of experimental analogue functional analyses.

Justification of Experimental Functional Analysis

Functional assessment methods aim to identify the variables that serve to evoke and maintain challenging behaviour. These methods may be informant-based, descriptive or experimental (analogue). In study one, indirect informant-based methods were adopted to examine the function of challenging behaviour associated with FXS and SMS. In the study reported in the current chapter experimental ABC experimental functional analysis methods were used to examine this same question. There are a number of advantages as well as disadvantages to experimental functional analysis and the following section aims to justify the use of these methods over and above alternative options.

Indirect functional assessment.

Indirect functional assessment methods involve interviewing significant others about the factors they believe to be related to the occurrence of challenging behaviour. Indirect measures do not involve the direct observation of challenging behaviour but instead measure the factors that informants report as being related to challenging behaviour.

A number of methods exist for conducting an indirect functional assessment. One means of conducting indirect assessments is to use semi-structured interviews, such as the *Functional Assessment Interview* (O'Neil, Horner, Albin, Storey, & Sprague, 1990). Other forms of indirect functional assessment, such as the *Motivation Assessment Scale* (MAS; Durand & Crimmins, 1988) or the *Questions About Behavioral Function scale* (QABF; Matson & Vollmer, 1995), involve the use of more structured interviews or checklists.

The advantage of indirect assessments lie in their ease of use and the low demands they place on the resources of the researcher or clinician. However, this is counterbalanced by genuine concerns surrounding the validity and reliability of such methods. Such measures provide an indirect measure of behaviour and measure only those variables that informants report as being responsible for its maintenance. Such reports are not necessarily valid or accurate, as demonstrated in a recent study (Hall, 2005). The threats to the internal validity of such instruments perhaps accounts for the poor psychometric data for measures such as the MAS (e.g., Sigafoos, Kerr, & Roberts, 1994). Even if respondents provide an accurate reflection of what they have observed the sample of observations on which their judgments are based may not be representative of the conditions that truly evoke and maintain the behaviour. In addition there are likely to be important inter-rater differences that influence the outcome of such measures (Sturmey, 1995; Vollmer & Smith, 1996). For this reason it has been recommended that instruments, such as the QABF, be used cojointly with other methods and to help inform and develop hypotheses which can be tested using alternative functional assessment methods (Hanley, Iwata, & McCord, 2003).

Descriptive functional assessment methods.

Descriptive assessments involve the identification of antecedent and consequent variables that are temporally related to the target behaviour and typically involve the direct observation of behavior in its natural settings. The conditional probability of certain events preceding or following the target behaviour can be calculated from such information using methods such as lag sequential analysis (Yoder, Short-Meyerson, & Tapp, 2004). Descriptive methods have been used to assess the function of a wide range of behavioural topographies such as aggression, bizarre speech, self-injurious behaviour, stereotypy and disruption (Lerman & Iwata, 1993).

As descriptive assessments include variables that are present in the natural setting in which the behaviour takes place, descriptive methods have been cited as being more ecologically valid than experimental methods (Hall, 2005). Such methods may be of particular use in identifying the influence of relatively idiosyncratic variables (Mace & Lalli, 1991). However this is not necessarily always the case and the presence of an observer alone has the potential to reduce the external validity of observations by virtue of reactivity effects. In addition adaptations can be made to increase the ecological validity of experimental methods so as to negate this apparent advantage.

There are a number of problems associated with descriptive methods. The demonstration of temporal associations between behaviour and environmental events is purely correlational and therefore falls short of the standards required to demonstrate a functional relationship. Without experimental manipulation or replication definitive conclusions regarding functional relationships are not possible (Bijou, Peterson, & Ault, 1968).

Due to the reliance on such correlations descriptive functional assessment methods may be especially vulnerable to both Type I and Type II errors. A particular concern is that irrelevant variables may mask functional relations due to their relative frequencies; that is there may be both a false positive and a false negative result (Iwata, Vollmer, & Zarcone, 1990; Oliver, 1991). For example, a behaviour maintained on an intermittent schedule of negative reinforcement may also be consistently correlated with social reprimands and disapproval. Even though such reprimands may exert no functional control over the behaviour, a descriptive assessment would falsely suggest that the behaviour is positively reinforced. Secondly, the variables that evoke and maintain challenging behaviour may be relatively idiosyncratic and be of such a nature as to escape the attention of the observer (Oliver, 1991). Finally, antecedents known by caregivers to evoke challenging behaviour may be deliberately avoided thereby precluding their assessment. Given the risk of making such Type I and Type II errors the use of descriptive assessments as the sole basis for treatment recommendations may in some circumstances be contraindicated (Lerman & Iwata, 1993).

Studies that have assessed the validity of descriptive assessments have typically used experimental analysis as the yardstick against which their validity can be compared.

Lerman and Iwata (1993) found direct naturalistic observations were unable to yield consistent ascriptions of function for five of the six subjects, suggesting that the validity of descriptive assessments may be limited. Likewise, Mace and Lalli (1991) reported that the descriptive assessment of bizarre speech suggested alternative functions than did experimental analogue methods. Other researchers have found agreement between analogue and descriptive assessments to be good when comparison is restricted to topographies for which both methods are able to ascribe function (Emerson, Thompson, Reeves, Henderson, & Robertson, 1995).

Recent developments have advanced descriptive methods and have in part addressed some of the disadvantages discussed above. One strategy to improve the utility of descriptive methods is to programme the occurrence of certain antecedent events that are known to be likely to evoke challenging behaviours. This method, known as a *structured descriptive assessment* (SDA), involves manipulating specific antecedent events, in a manner similar to those used in analogue settings, whilst observing naturally occurring consequences. SDAs have been shown to represent an improvement over standard descriptive assessment methods (Anderson & Long, 2002; Freeman, Anderson, & Scotti, 2000).

Anderson and Long (2002) describe several advantages to the SDA over alternative functional assessment methodologies: 1) unlike analogue assessments it is not necessary to remove individuals from their natural environment or disrupt their natural routine; 2) because these assessments occur in the natural environment they may be more ecologically valid than analogue methods; 3) unlike standard descriptive methods SDAs ensure that relevant antecedent events are delivered at a rate similar to those found in analogue conditions, reducing the likelihood of a missed detection. However, many of the weaknesses associated with descriptive assessments remain intact with SDAs. For example, behaviours that are maintained on a thin schedule of reinforcement may be relatively difficult to identify.

Experimental methods.

Experimental analogue methods involve the contrived manipulation of environmental variables to establish relations between the experimental manipulation and behaviour. As environmental events are experimentally controlled such methods provide a more convincing demonstration of the determinants of challenging behaviour.

Analogue methods are an umbrella term, covering two different models of functional analysis. The first model was initially described in a seminal paper by Iwata et al (1982/1994) and has been termed the ABC model of functional analysis (Hanley et al., 2003). Typically this method involves the repeated presentation of several experimental conditions, in which putative antecedents (i.e., discriminative stimuli and motivating operations) and consequences (i.e., reinforcers) are systematically manipulated using a multi-element design. The experimental conditions originally reported by Iwata et al included a *social disapproval* condition, a *social demand* condition, an *alone* condition, and a control *play* condition (see chapter one for a detailed description of these conditions).

Several modifications of this design have been used over the past twenty five years. For example, *tangible* (Day, Horner, & O'Neill, 1994) or *social avoidance* (Hagopian, Wilson, & Wilder, 2001) conditions have been added to test additional hypotheses about the function of challenging behaviours. The procedure and experimental design have been adapted in a number of ways to accomodate time restraints and other practicalities (Iwata & Dozier, 2008; Northup et al., 1991). Other researchers have simply shortened the duration of sessions from 15-min to either 10-min or 5-min conditions (Hanley et al., 2003).

An alternative 'analogue' model was reported by Carr and Durand (1985). This model, termed the AB model, involves manipulating the amount of antecedent attention (i.e., between either 100% of intervals or 33% of intervals) and the difficulty of tasks (i.e., between either easy or difficult). This results in three experimental conditions (i.e., easy 100, easy 33, and difficult 100), in which the easy 100 condition is designed to act as a control against which any changes in behaviour in the other conditions can be compared. Challenging behaviours are typically ignored, unless the child has eloped, whereby the experimenter leads the child back to their seat, or the behaviour poses a physical risk, whereby the child is briefly restrained.

General critique of analogue methodology.

There are a number of critiques that have been made about the use of analogue methodology in general. Such critiques typically focus on the ethical concerns associated with such work and the issue of external validity.

One of the most significant ethical criticisms directed at analogue methods has been that such methods are explicitly designed to evoke challenging behaviour (Hastings & Noone, 2005). When dealing with behaviours, such as self-injury, that have the potential to incur injury to the individual this is a clear ethical concern. This concern appears to have been one of the greatest barriers to the use of analogue methods.

The risk of injury can to some extent be attenuated by making practical adjustments to analogue sessions. For example, Iwata et al (1982/1994) describe clear criteria for session termination based on the frequency, intensity or duration of self-injury, and the use of medical staff to help in the event of injury. In addition protective equipment can also be incorporated into analogue assessments (O'Reilly, Murray, Lancioni, Sigafoos, & Lacey, 2003).

There also exists an ethical case for the use of analogue methods over their descriptive or indirect counterparts. For example, individuals have a right to the most effective treatment procedures available (Van Houten et al., 1988). If, as has been argued, analogue methods may be more able to deal with the threats to internal validity posed by Type I or II errors, treatments based on the results of a functional analysis are more likely to be effective than alternative methods. Likewise given that treatments based on an understanding of the function of challenging behaviour are likely to be less restrictive (Pelios, Morren, Tesch, & Axelrod, 1999) there may be a case for the use of analogue methods to ensure that the individual receives the least restrictive treatment option available.

There are legitimate concerns surrounding the resource intensive nature of functional analysis methods. For example it has been estimated that the abbreviated Northup et al. procedure consist of 54-68 'in session' minutes; whilst the extended Iwata et al. procedure consists of 270-340 'in session' minutes and may take 1-2 weeks to complete (Matson & Minshawi, 2007). This has been used as a justification for seeking alternative, less time-consuming means of assessing challenging behaviour. However, as Vollmer and Smith (1996) note, time constraints alone are not defensible objections to the use of functional analysis.

It has been suggested that analogue methods may not be appropriate for certain topographies of behaviour, particularly for low-rate, high-intensity behaviours, such as some forms of aggression, fire setting, and inappropriate sexual behaviour (e.g., Whitaker, 1993). For example, some behaviours may occur for durations longer than the 10-15 minutes involved in functional analysis sessions or may occur only at very low rates (i.e., only once a week). In some cases the severity of behaviour may preclude any extended period in which the person is able to freely engage in the behaviour. Whilst careful consideration is required one particular benefit of analogue methodology is its flexibility and with appropriate modifications such topographies of behaviour have been included in analogue functional analyses (e.g., O'Reilly et al., 2003). In addition, the functional analysis of pre-cursor behaviours has been shown to be an effective means of identifying the function of high-intensity challenging behaviours that are part of the same response class (R. G. Smith & Churchill, 2002).

Given that analogue conditions aim to provide an approximation of the variables that maintain behaviour in naturalistic settings the issue of external validity is of some importance. Some have questioned the validity of experimental analysis methods. Indeed, as Mace states, "*experimental analyses...may not generalise outside the analog conditions*" (Mace, 1994). The use of indirect and descriptive assessment methods can help in the identification of relatively idiosyncratic antecedent and consequent manipulations to incorporate into an analogue analysis (Mace, Lalli, Lalli, & Shea, 1993), increasing both the external and internal validity of analogue methods. Therefore, it is not necessarily the case that analogue methods have lower external validity than alternative modes of functional assessment. Indeed, Iwata and Dozier (2008) suggest that the results of numerous treatment studies suggest that the contingencies provided in a functional analysis are 'close enough' to those found in the natural environment to form the basis of effective treatments (p. 4).

Serial dependency between sessions poses a potential problem for any design that involves the repeated measurement of behaviour over time (Sturmey, 1995). This is particularly true for analogue methods, which involve rapid alterations between different experimental conditions. In the current context, for example, an attention condition preceded by a no interaction condition may result in a very different pattern of responding than an attention condition that is preceded by a demand condition. Whilst a potential confound such factors are not beyond experimental control. In addition to randomising the order in which conditions are implemented, within-session analyses can be conducted to monitor potential order effects. The flexibility of analogue methodology facilitates the control of such confounds once detected. Iwata et al (1994) recommend implementing functional analysis conditions in a 'semi-random' manner in order to reduce the extent that preceding conditions function as MOs for those that follow. Such a semi-random schedule would, for example, involve a no interaction condition never following an attention condition to reduce the likelihood of an extinction burst for attention maintained behaviour.

Benefits and costs of the ABC model of functional analysis.

The AB model of functional analysis provides a less rigorous demonstration of the function served by challenging behaviour, as potential sources of reinforcement are not manipulated in such analyses (Hanley, Iwata & McCord, 2003). Such methods may lead to erroneous conclusions about the function served by certain behaviours due to this reduction in experimental control. This contrasts against the ABC model of functional analysis whereby experimental control is demonstrated over the behaviour-consequence contingency. Some recent evidence exists to suggest that Type II errors may be more likely to occur when using AB assessments in comparison to their ABC counterparts (Potoczak, Carr, & Michael, 2007; Worsdell, Iwata, Conners, Kahng, & Thompson, 2000). There may also be some specific ethical concerns regarding the use of AB assessments. The 'consequence' in such assessments is to simply ignore challenging behaviour, for behaviours maintained by attention or escape this is akin to placing the response on extinction whilst the EO for the maintaining variable remains in effect. Recent evidence has shown that such manipulations are likely to evoke high levels of challenging behaviour (O'Reilly et al., 2006) and one would expect there to be an increase in the intensity of such behaviours as a result of an extinction burst (Vollmer et al., 1998). Further research is needed in this area but AB assessments may be associated with more costs from an ethical perspective, than is commonly acknowledged.

There are some ethical and practical concerns specifically associated with the ABC model which require some consideration.

A common criticism to have been directed at ABC analogue methods is that exposure to ABC contingencies within a functional analysis may over time lead to the acquisition of a new behavioural function (Mace, Lalli, & Lalli, 1991); that is either a previously uncommitted behaviour may acquire operant status, or the function of an existing operant may change as a direct result of the assessment procedure. Iwata, Vollmer and Zarcone (1990) acknowledge that the ABC model may establish new relations that have the potential to generalise to the natural environment. They recommend curtailing analogue assessments to the shortest time required in order to demonstrate functional control as a means of minimizing such potential effects. Despite such concerns it has been suggested that the probability of this occurring in experimental conditions is no more than and may be less than in naturalistic conditions (Iwata et al., 1982/1994).

During an analogue session several non-target behaviours may occur and experimenter responses to such behaviours may function as a potential threat to internal validity (Sturmey, 1995). Iwata et al recommend that non-target behaviours be ignored. However, if non-target behaviours form part of the same response class as the target behaviour, then their extinction may lead to increases in the target behaviour (Sprague & Horner, 1992). Alterations in the pattern of responding for the target behaviour would in such circumstances be an artefact of the effects of extinction on other members of the response class. To overcome such problems non-target behaviours need to be monitored to determine whether they do indeed form part of the same response class as target behaviours, when they do all members of that response class should be included in the assessment (Sturmey, 1995). Hanley, Iwata and McCord (2003) suggest that best practice would be to minimise the number of different topographies of challenging behaviour included in a single functional analysis and if multiple topographies are to be included that each topographical class should either be subjected to extinction or separate topographies should be graphically analysed before being aggregated into a single data path.

There is considerable latitude in the design and implementation of analogue conditions, and as such there may be an erroneous ascription of function to certain target behaviours. For example, there appears to be several aspects of task demands that evoke challenging behaviour (R. G. Smith, Iwata, Goh, & Shore, 1995) and for some individuals the high level of social interaction in the play condition may act as an aversive stimulus (Hagopian et al., 2001). High rates of challenging behaviour in this condition may lead to the false inference that the behaviour is automatically reinforced; when in fact the behaviour is negatively reinforced (by the removal of attention due to the DRO component embedded within the analogue condition). Similarly the alone condition may not be truly implementable (i.e., in cases in which there is no one-way screen) and stimuli that are discriminative for attention may remain in place in the modified no interaction condition (Hanley, Iwata & McCord, 2003). The social contact provided in the social disapproval condition may in some circumstances serve to punish and not reinforce attention-maintained behaviour (Fisher et al., 1994). Such concerns are not beyond experimental control and it appears that a particular strength of analogue methodology is in its ability to demonstrate functional relationships between relatively idiosyncratic events and operant behaviour.

Given that ABC analogues are the benchmark against which other methods are compared the issue of reliability is of some importance. Findings in this regard have been somewhat equivocal. Martin, Gaffan and Williams (1999) reported a high percentage of undifferentiated patterns of responding and weak to poor test-retest reliability using experimental analogue methods. However, the use of brief assessment conditions that were 5-min length, and the absence of any contingencies, apart from in the attention condition, may have hindered discrimination between conditions for participants in the study. Toogood and Timlin (1996) report that experimental methods failed to ascribe function in 50% of cases compared with 11-15% for clinical interview and MAS (Durand & Crimmins, 1988) and 18% for descriptive analysis. This contrasts markedly with the 95% success rate reported in an epidemiological study by Iwata et al (1994) although different decision-making criteria were used in each study in order to ascribe function. There does appear to be some relationship between the length of assessments and the likelihood of ascribing function. For example, Derby et al (1992) report that brief functional analyses involving some 79 people were only successful in recording the function of behaviour in 50% of cases. Hanley, Iwata and McCord (2003) recommend that researchers and clinicians should continue to work to identify the sources of variability that give rise to 'noisy' functional analysis results.

In sum, the strength of ABC analogue assessments lies in the level of experimental control that such methods provide. Such methods utilise the advantages of experimental single-case design to identify the variables of which behaviour is a function. Whilst concerns exist about the appropriateness of using such methods it does seem that their advantages outweigh the disadvantages when judged against indirect, descriptive and AB analogue alternatives. In addition, it is far from clear that one method of analogue assessment holds any advantage over the other in terms of their ethical implications.

The study reported in the current chapter adopted ABC functional analysis methods in order to examine within- and between-group differences in behavioural function between a small number of children with FXS and SMS. Individual variations in challenging behaviour were examined across several different environmental conditions. To the author's knowledge this is the first study to conduct such a between-group comparison using experimental analogue methods. To address some of the ethical concerns that exist regarding the acceptability of analogue methods (Hastings & Noone, 2005), data were collected on the social acceptability of the assessment (Wolf, 1978).

Method

Participants and Ethical Considerations

All participants had a confirmed diagnosis of either FXS or SMS and were selected primarily according to their geographical proximity to Canterbury. The majority of participants were located within a 150 mile drive of the study base. Participants from other geographical areas were then recruited. No family who had been contacted via NHS Regional Genetics Centres in the study reported in the previous chapter (henceforth referred to study one) were approached about the current study (henceforth referred to as study two). Otherwise the inclusion and exclusion criteria remained the same as in study one. There were a total of 8 children in the FXS group and 6 in the SMS group who took part in the current study.

The study received ethical approval from the Tizard Centre Ethics Committee. As participants were expected to display challenging behaviour during the course of study two it was important that a rigorous criterion (expressed in terms of either degree of injury or level of responding or both) was established for the termination of experimental sessions. This criterion was set following consultation with the child's parents, and carers. If a participant's target behaviour met criterion then the session was to be halted and a decision made as to whether the session should be either postponed or resumed. In no cases did any participant's behaviour reach the criteria for session termination. Inclusion in the study was under continual review and regular discussions were held between the author, parents and other relevant individuals as to the individual's continued participation in the study. In addition regular discussions regarding each participant were held in supervision sessions. It was felt that the risks to which participants were exposed were no greater (and in some cases considerably less) than those found in the natural environment.

Response Measurement and Reliability

Target behaviours included *stereotypical behaviour*, *self-injurious behaviour*, *destructive behaviours*, *non-compliance* and *aggressive behaviours*. Response definitions were developed on an individual basis and were based on responses made in study one, as well as informal observations and further discussion with parents and carers. In some cases, either unexpected behaviours occurred during the functional analysis or there were no occurrences of expected target behaviours. In cases where behaviours occurred that were not initially reported by parents/caregivers then where possible these were included for analysis. In cases where target behaviours did not occur during the functional analysis this is noted in the discussion of the specific individual's results below. All non-target behaviours were ignored, unless the child attempted to leave the assessment area, in which case the child was redirected to the area using minimal amounts of interaction.

All sessions were videotaped to facilitate data collection and reliability checks. A 10-s partial-interval method was used to code the data for all participants. All responses were scored using pen and paper. Data were then inputted into *Excel* ™ using a desktop computer.

A second observer recorded participant behaviour for at least 29% of functional analysis conditions for each participant. All target behaviours were collapsed and coded according to their response class (see individual participant descriptions). The percentage of inter observer agreement was calculated using the exact agreement method. Agreement was defined as both observers agreeing on the occurrence and/or non-occurrence of the target behaviour(s) in any given interval. Disagreement was scored as any discrepancy between the observers in any given interval. The number of agreements was divided by the number of agreements plus disagreements, and the resulting value was multiplied by 100%. Overall inter-rater reliability for all intervals (R-tot), as well as the reliability for only the occurrence (R-occ) and the non-occurrence (R-non occ) was calculated for each response class. In each case the most stringent measure is presented for each participant in the text below. In addition the Kappa statistic was calculated for each response class. Appendix 5 shows all IOA data for all participants and response classes. For individuals where the IOA was unacceptably low (less than 50%) the second observer was given additional training and asked to recode those topographies for which there was a problem. The dissertation supervisor reviewed all cases where additional training was required.

Experimental Design

Experimental conditions were implemented using a multi-element design. Participants were exposed to up to six different experimental conditions, which were implemented in a randomised fashion.

General Procedure

A subset of families who had taken part in study one and indicated a willingness to take part in study two were sent an information sheet and consent form about taking part in study two. One child who took part in study two (Shelley) was recruited via other means. Families were asked to return a completed consent form if they were interested in taking part in the second study. Families were then contacted to establish a convenient time and date for the researcher to visit.

Before beginning the study there was an initial visit in which key stakeholders (families and/or teachers) were talked through the procedure involved in conducting functional analysis. A DVD depicting the assessment methods to be used was also shown on request. At this initial meeting response definitions were drafted for each child and preferred tangibles were identified for use in the *Tangible* condition. Other factors that may have influenced the results of analogue assessments were also identified (i.e., type of attention provided, presence of any health conditions). In some cases this led to changes in the design of the specific analogue conditions described below. Some time was also spent interacting with each child on this initial visit.

There was a maximum of four subsequent visits to either the child's home or school in order to run the functional analysis and each visit lasted up to a maximum of 90 minutes. Functional analyses are often conducted in specially designed rooms, equipped with one-way mirrors, to minimise the influence of extraneous variables and to facilitate the running of certain conditions (Iwata et al., 1982/1994). As the analyses were conducted some way from the study base (Canterbury, UK), this was not possible in the current study, and assessments were conducted in a room in the participant's home or school. In some cases, due to the structure of the room in which the analyses were conducted, it was not possible to run particular conditions without the child eloping from the room and/or interacting with caregivers. In such cases this specific condition was removed from the analysis. Where this occurred this is noted in the discussion of individual results.

Each participant was exposed to a maximum of six different experimental conditions. Each session lasted 5-min.

A brief description of each condition is provided below. In cases where modifications were required to specific conditions these are highlighted in the discussion of individual participants (Sean, John, Patrick and Matt) in the results section below. Modifications were only made to conditions if the results of the QABF suggested that the individual displayed challenging behaviour that served a certain function. For example, the attention condition was only modified if the results of the QABF suggested that the individual displayed attention-maintained behaviour (indicated by a score of 10 or more across one or more topographies).

1) *Attention.* The participant was asked to play with some toys. The experimenter then pretended to read a book and stated that he was going to do some work. Attention was given for 10s contingent on each occurrence of challenging behaviour and took the form of statements of concern and mild disapproval paired with non-punitive physical contact, whilst the child was redirected to his toys. All other responses were ignored. This condition aimed to test whether attention served to maintain each child's challenging behaviour.
2) *Academic demand.* Educational activities were selected following discussion with informants and direct observation. Completion of tasks was judged to have a low probability of occurrence and never occurred spontaneously. Learning trials were presented to the participant using a three-prompt procedure (verbal request, modelling,

and physical guidance). If the child did not respond to the demand after 5 seconds the experimenter gave a gestural or model prompt indicating the correct response. If the participant still did not respond a physical prompt was used. Social praise was given contingent on the successful completion of the learning trial, except on those occasions in which physical guidance was required for task completion. Academic demands were removed for 10-s contingent on target behaviours. This condition aimed to test whether each child's challenging behaviour was negatively reinforced by the removal of academic demands.

3) *No interaction.* The experimenter turned his back to the child and provided no interaction contingent on any behaviour. The child had no access to toys. This condition aimed to test whether target behaviours were maintained by their non-social consequences.
4) *Unstructured play.* The child had access to preferred toys, whilst the experimenter delivered social praise at least every 15-s following the first 5-s period in which challenging behaviours had not occurred. All target behaviours were ignored and the experimenter provided no demands on the child. This condition aimed to provide a control condition and provide an analogue of the 'enriched environment'.
5) *Social avoidance.* The child was provided with preferred toys and asked to play with them. The experimenter provided continuous attention by talking with the child and commentating on their play. If challenging behaviours occurred then attention was removed for approximately 10-s (c.f., Hagopian et al., 2001). This aimed to determine whether the child's challenging behaviour was maintained by the removal of social attention.
6) *Tangible.* In the tangible condition, toys or food items, identified as highly preferred, were placed in sight but out of reach of the child. The experimenter delivered the

tangible for approximately 10s contingent on the occurrence of target behaviours or the child was given access to food on a FR-1 schedule.

At the end of the assessment families were sent a copy of an assessment based on the results of the analogue assessment together with some basic recommendations. Key stakeholders (parents and teachers/therapists) were asked to return an anonymised questionnaire in which the acceptability of the assessment was rated. The questionnaire was an amended version of the shortened Treatment Acceptability Rating Form-Revised (Reimers, Wacker, Cooper, & DeRaad, 1992) and included 9 items each of which was rated using a 5 pt rating scale (1 = strongly disagree – 5 = strongly agree).

Data Analysis

In order to overcome some of the problems of relying solely upon visual analysis a structured method was used to ascribe behavioural function. A number of different methods have been proposed to formalise this process (Hagopian et al., 1997; Martin et al., 1999; Toogood & Timlin, 1996).

In the current study, a modified version (see Martin et al., 1999) of the Hagopian et al criterion for differentiation was used. One of the relative advantages of the Hagopian et al method is the ability to deal with relatively complex patterns of data (Hanley et al., 2003). Other methods, such as the probability based Criterion Z approach proposed by Martin et al (1999) or that proposed by Toogood and Timlin (1996) have no clear criteria as to how to interpret such data. In addition, due to the relatively low number of replications of each experimental condition the probability-based approach proposed by Martin et al was not deemed appropriate for the current study.

Using the modified Hagopian et al method a condition is considered differentiated when at least 50% of the data points for one condition falls 1 standard deviation (SD) above the mean of the play condition. A criterion line (CL) is drawn at this mark on the graph of the functional analysis[26]. In situations where levels of responding in the play condition are near zero the criterion is set at a minimum of .5 responses per minute (in the current study this would mean a CL being set at a minimum of 8.33% of intervals). The CL is marked on all functional analysis graphs presented in the individual results section below, apart from for one participant (Katie) where the response occurred at an especially low rate. Hagopian et al also propose criteria for the interpretation of unusual data paths. In cases where these additional criteria were used this is noted in the results section below and the criteria used are presented as a footnote.

In order to ensure that the method of interpretation was consistently applied all judgments were made initially by the first author and checked for consistency by the first author's dissertation supervisor. Any disagreements were re-analysed and re-checked for consistency.

Results

Analyses and resulting conclusions are presented for each individual who took part in the current study[27]. Summary data for each syndrome group are also presented. The primary role of the latter was to examine the hypothesis that there were within- and between-group differences in socially-mediated functions of challenging behaviour. As such, response classes that served a sole automatic function were excluded from the summary data for each syndrome group.

Participants with FXS. Individual Functional Analyses

A total of eight individuals with a diagnosis of FXS took part in study two. All participants were male and their parents had taken part in study one. Table 5.1 provides some basic demographic information for each participant.

[26] A condition is considered differentiated when 50% or more of data points fall at or above the CL.

[27] A table depicting the correspondence between each individual's results for indirect and analogue assessment methodologies is presented in Appendix 6.

Given the small numbers involved in the current study it is possible that any within- or between-group differences in behavioural function could be due to sampling bias. Categorical data from the QABF for the 8 children with FXS were compared against the remainder of the FXS group from study one. A function was deemed to be present if the individual scored 10 or more for a given subscale. Using a significance level of $p = .05$, a series of chi-square analyses revealed no significant differences between the two sub groups for any scale across any topography. This suggests that the participants with FXS were representative of those who took part in study one in terms of the potential function served by challenging behaviours.

Table 5.1

Characteristics of Participants with FXS.

Participant	Age*	Total ABC	Age equivalent (Vineland Sub-domains)		
			Communication	Daily Living	Socialization
Abe	9yrs 8mths	108	1yr 9mths	1yrs	2yrs
Greg	9yrs 8mths	112	3yrs 1mth	2yrs	2yr 1mth
Jacob	8yrs	51	4yrs 1mth	4yrs2mths	5yrs 5mths
Luke	15yrs 10mths	59	1yr 3mths	1yr 1mth	0yrs 6mths
Theo	10yrs 4mths	39	3yrs 1mth	2yrs 8mths	6yrs 9mths
John	13yrs	128	2yrs 2mths	2yrs 3mths	3yrs 1mth
Calum	15yrs 1mth	80	5yrs 3mths	8yrs 4mths	5yrs 7mths
Richard	11yrs 7mths	47	5yrs 5mths	3yrs 1mth	4yrs 10mths

*Age at the beginning of study two

Abe.

Abe was nine years old and had a diagnosis of FXS and autism; he attended a school for children with severe learning disabilities and lived in the family home. Abe would communicate using one or two word utterances or by signing. The QABF was completed as part of study one by Abe's mum, which indicated that challenging behaviours appeared to serve multiple functions, with notably high scores on the tangible and demand subscales. The total subscale scores for Abe are presented in Table 5.2.

Target behaviours included self-injurious behaviour (finger-biting with accompanying low guttural vocalisation, and hand-biting), aggression (kicking, pulled punch, slapping, and pinching), and property destruction (throwing toys). Target behaviours were collapsed into either finger-biting or other challenging behaviours. Inter-rater reliability was calculated for 41.6% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for finger-biting was 92.1% and for other challenging behaviours was 87.5%.

Table 5.2

Abe. Total subscale scores from QABF subscales

	Self-injurious behaviour	Aggression	Property Destruction
Abe.			
Attention	7	7	N/A
Tangible	15	15	N/A
Demand	12	15	N/A
Physical discomfort		10	10
N/A			
Automatic	12	2	N/A

All experimental sessions were conducted in Abe's bedroom. Academic tasks used in the demand condition were part of Abe's current educational goals (matching to sample, counting) and were tasks which he had not yet achieved mastery of. The tangible condition involved withholding access to a preferred toy, which was identified via parental report. Due to practical difficulties associated with conducting the analysis in Abe's bedroom, (he would close the door, thereby making direct observation impossible) a no interaction condition could not be included. All other experimental conditions were run in accordance with the protocol for the study.

Results of the functional analysis suggested that challenging behaviour displayed by Abe formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.1. Abe. Percentage of intervals with finger biting during functional analysis.

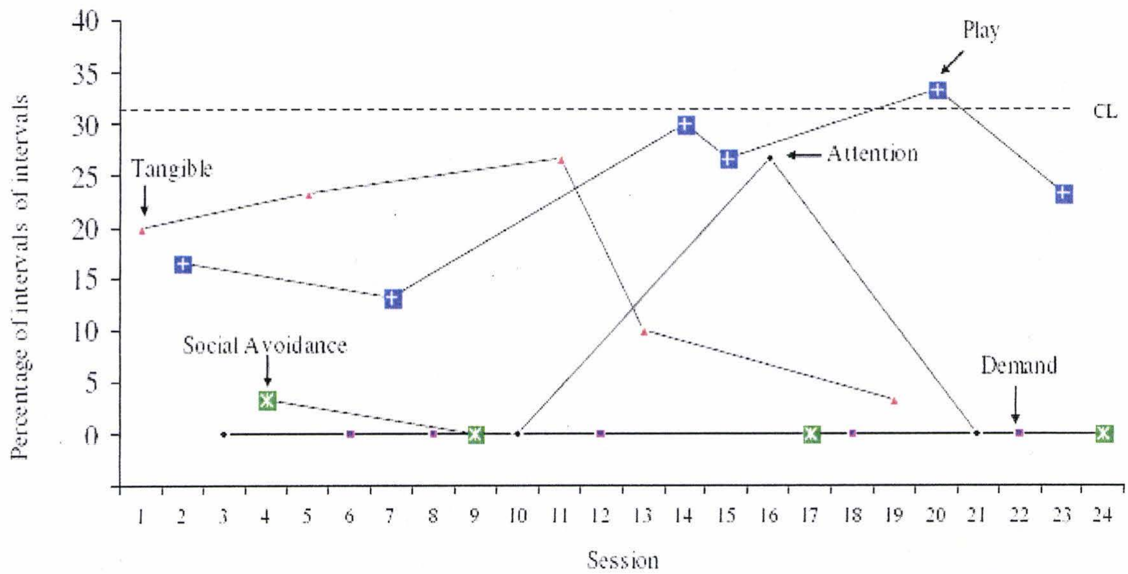


Figure 5.1 shows session by session data for the occurrence of finger-biting. There were elevated rates of finger-biting in the play condition (M = 23.9; range = 13.3 – 33.3) and to a lesser extent the tangible condition (M = 16.6; range = 3.3 – 26.6). There were also high rates of finger-biting in a single session of the attention condition (M = 6.7; range = 0 – 26.6). There was a single occurrence of finger biting in the social avoidance condition (M = .83; range 0 - 3.3) and zero occurrences of finger-biting in the demand condition of the functional analysis.

Due to the high level of challenging behaviour occurring in the play condition no condition met the criteria for differentiation between experimental conditions (Hagopian et al., 1997). This suggested that further analysis was merited to explore a potential automatically reinforced function.

Due to the unclear pattern of responding for finger-biting an exploratory analysis of the data was conducted to test the hypothesis that finger-biting was occasioned by the presence of specific stimuli. It appeared that Abe displayed finger-biting more frequently in sessions in which he interacted with certain preferred toys that shared similar characteristics (i.e., produced movement and sound), such as a toy train, balloons and a penguin toy. The percentage of intervals in which Abe interacted with these stimuli is shown in Figure 5.2, along with the percentage of intervals in which finger-biting occurred. Finger-biting appears to have been more likely to occur in sessions in which Abe had access and interacted with these toys than in sessions in which there was no toy interaction[28]. Indeed, apart from in session 4, there were no occurrences of finger-biting in the absence of interaction with these stimuli.

Figure 5.2. Abe. Percentage of intervals with toy interaction and finger biting across all conditions

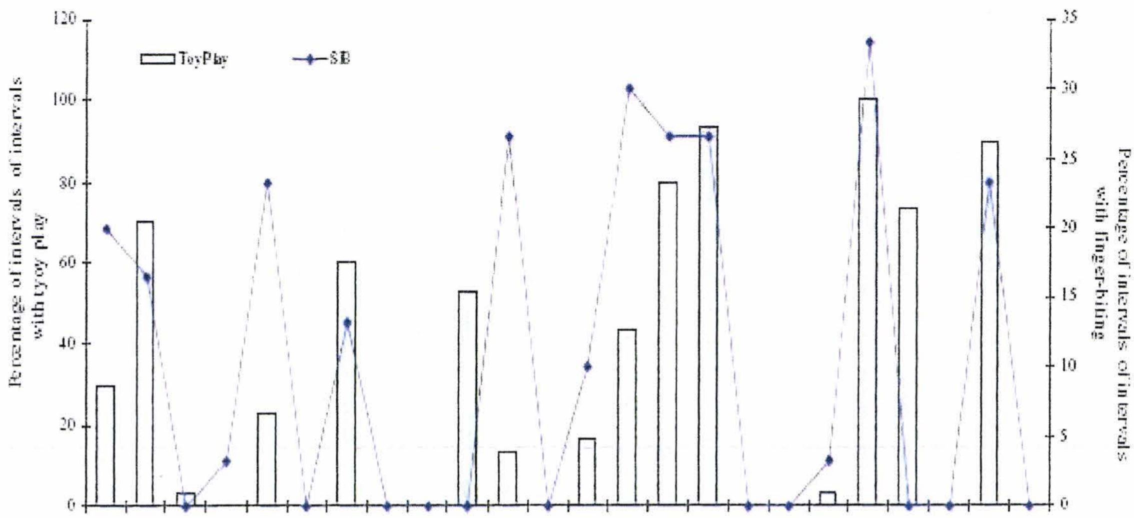
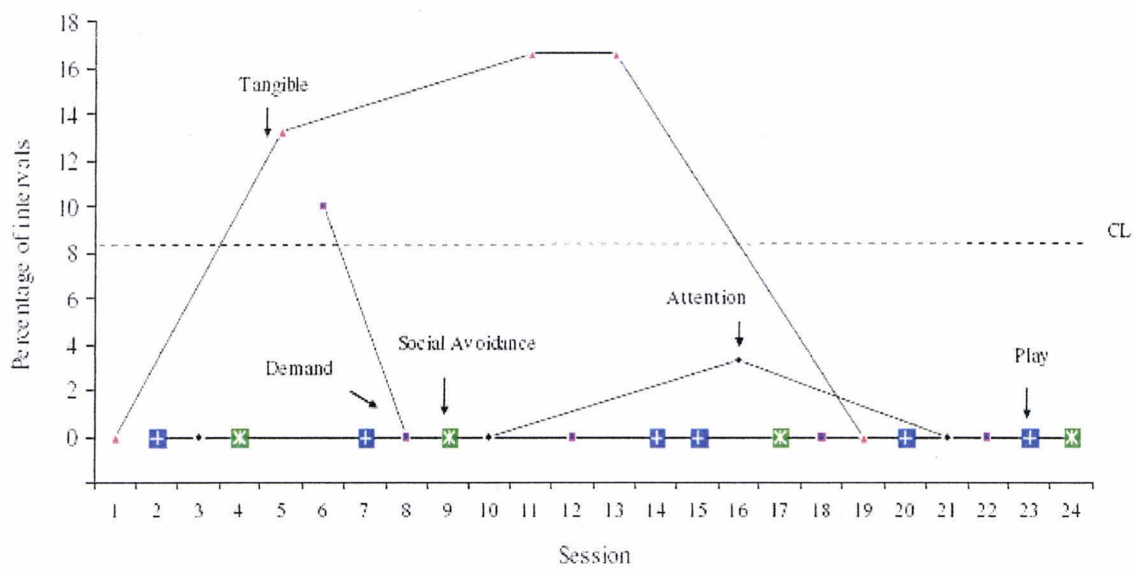


Figure 5.3 shows session by session data for the occurrence of all other topographies of challenging behaviour, excluding finger-biting. There were heightened

[28] The low levels of finger-biting occurring in session 21 may have been due to the fact that Abe had a pacifier in his mouth throughout the session. As such the target behaviour could not occur. Anecdotally, Abe brought his finger to his mouth throughout the session without it making contact with his teeth.

rates of challenging behaviour in the tangible condition of the functional analysis (M= 9.3; range= 0 - 16.6). There were also heightened levels of challenging behaviours in the initial demand condition of the functional analysis, although this reduced to zero rates in all subsequent demand sessions (M = 2; range = 0 - 10) . There were near zero levels of challenging behaviour in all attention (M= .83; range = 0 – 3.3), play (M= 0; range = 0) and social avoidance (M= 0; range = 0) conditions.

Figure 5.3. Abe. Percentage of intervals with other challenging behaviours during functional analysis.



As at least 50% of scores for the tangible condition lay above the CL the data met the criteria for differentiation suggesting the behaviour was tangible-maintained.

In sum, data for Abe suggested that finger-biting was automatically reinforced and some evidence was provided to suggest that it was occasioned by the presence as opposed to absence of particular stimuli, a pattern of responding that corresponds to that reported in other studies (Friman, 2000; Van Camp et al., 2000). Other topographies of challenging behaviour appeared to form an alternative response class and were maintained by access to tangibles. Anecdotally, parents reported that Abe frequently displayed challenging behaviour in the presence of demands. This was directly observed when ABA therapists presented academic demands to Abe. However, when the lead researcher presented the same demands using the same prompts, apart from in the initial demand condition, Abe displayed no challenging behaviour. It may be that this was due to differences in the discriminative properties associated with each individual or that some aspect of the lead researcher's behaviour attenuated the aversiveness of the demand. Alternatively, challenging behaviour that occurred in the context of demands in the natural setting may have been associated with the deprivation of tangibles and thereby evoked tangible-maintained behaviour (as a result of the behaviour-altering effect of the MO) irrespective of the demand per se.

Greg.

Greg was nine years old and had a diagnosis of FXS. Greg communicated by one or two word utterances; he attended a school for children with severe learning disabilities and lived in the family home. The QABF was completed as part of study one by Greg's mum, which indicated that Greg displayed escape maintained aggressive behaviour and automatically reinforced property destruction. The total subscale scores for Greg are presented in Table 5.3 for each topographical category.

Table 5.3

Greg. Total Subscale Scores from QABF Subscales

	Self-injurious behaviour	Aggression	Property Destruction
Greg.			
Attention	N/A	1	0
Tangible	N/A	5	2
Demand	N/A	14	2
Physical discomfort	N/A	6	0
Automatic	N/A	0	14

Target behaviours for Greg included; aggression (hitting the therapist on the head with an object), spitting, and property destruction (banging objects repeatedly whilst crying). Target behaviours were collapsed into either hitting the therapist on the head or other challenging behaviours. Inter-rater reliability was calculated for 38.1% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for head hitting was 100% and for other challenging behaviours was 95.4%.

All experimental sessions were conducted in the lounge area of Greg's home. Academic tasks used in the demand condition were selected following initial assessment and involved a matching to sample task. The tangible condition involved withholding access to the television. Only a single no interaction condition was included in the experimental analysis as the layout of the room made it difficult to contain Greg in the designated area and he would frequently leave the assessment area to enter the kitchen and speak with his mother.

Results of the functional analysis suggested that challenging behaviour formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.4. Greg. Percentage of intervals with head hitting during functional analysis.

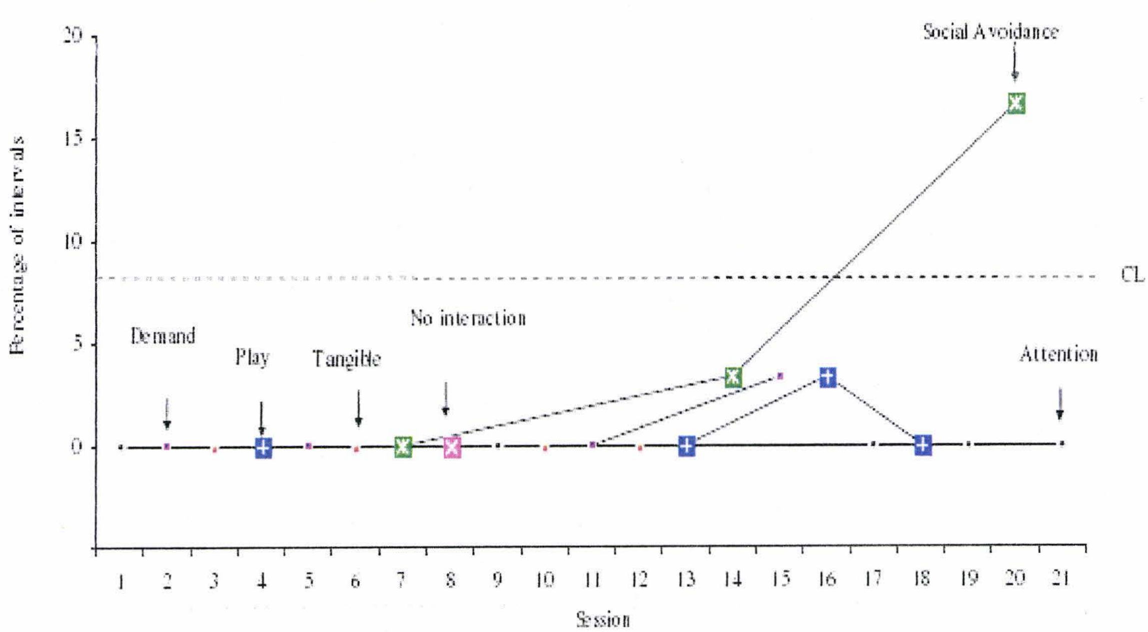
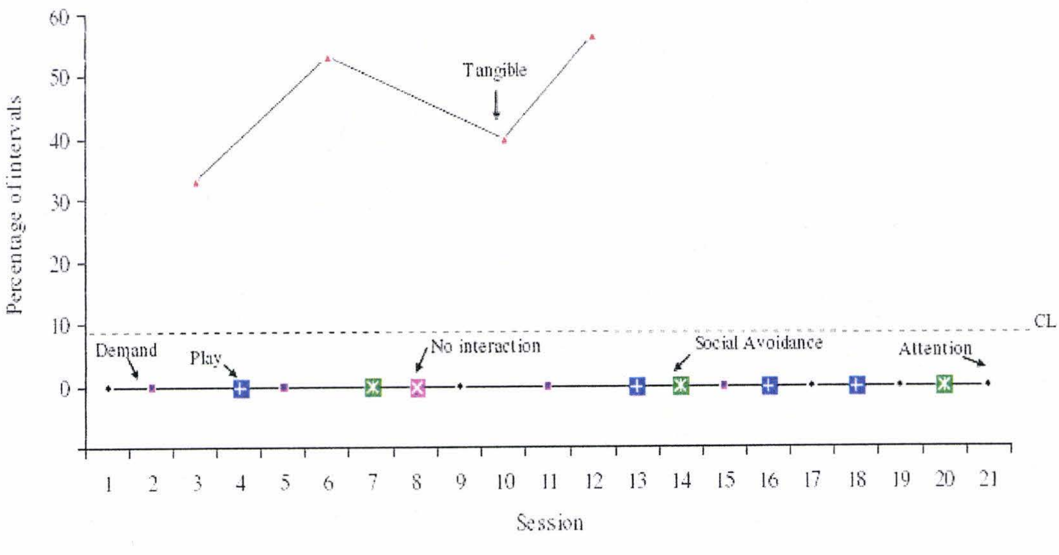


Figure 5.4 shows session by session data for the occurrence of head hitting. There were elevated rates of head hitting in the social avoidance condition (M = 6.63; range = 0 - 16.6) and to a lesser extent the play condition (M = .83; range = 0 – 3.3) and demand condition (M = .83; range = 0 – 3.3) of the functional analysis. There were zero occurrences of head hitting in attention, tangible and no interaction conditions.

As head hitting occurred at a relatively low rate, the criteria suggested by Hagopian et al (1997) for such behavioural patterns was applied to ascribe function[29]. Using this criterion, there was differentiation in the social avoidance condition of the analysis, suggesting that the behaviour may have been maintained by escape from social attention. Further support for this hypothesis is provided by the fact that the only other conditions in which the response occurred (play and demand) are characterised by relatively high levels of social contact.

Figure 5.5. Greg. Percentage of intervals with challenging behaviour (excluding aggression) during functional analysis.



[29] "In cases in which most of the data points are low, the condition in which all or most of the higher rate behavior occurs is considered to be differentiated.... However, one of those high points must occur in the last half of the assessment." (see Hagopian et al., 1997, p. 325).

For Greg, there were a total of 6 intervals in the social avoidance condition in which head hitting occurred, in comparison to 1 interval in the play condition and 1 interval in the demand condition.

Figure 5.5 shows session by session data for the occurrence of other challenging behaviours (property destruction and crying; spitting). There were elevated rates of challenging behaviours solely in the tangible condition (M = 45.8; range = 33.3 – 53.3). There were zero occurrences of challenging behaviour in any other condition.

As at least 50% of scores for the tangible condition lay above the CL the data met the criteria for tangible-maintained challenging behaviour.

Data for Greg suggested that head-hitting may have been negatively reinforced by the removal of social attention. Other topographies of challenging behaviour appeared to form an alternative response class and were tangible-maintained. Anecdotally, Greg's parents reported that he was also especially likely to display challenging behaviour when out of the house and asked to do things that were novel (e.g., going somewhere new). An analogue of this situation could not be included in the functional analysis for practical reasons, however.

Jacob.

Jacob was eight years old and had a diagnosis of FXS. Jacob was verbal using simple sentences to communicate; he attended a school for children with moderate learning disabilities and lived in the family home. The QABF was completed as part of study one by Jacob's mum, which indicated that Jacob displayed tangible-maintained challenging behaviour. The total subscale scores for Jacob are presented in Table 5.4.

Target behaviours for Jacob included self-injury (slapping his hand against his head, forcing his forehead against surfaces, arm-biting), aggression (hitting others) and property destruction (banging objects, throwing items, kicking surfaces). There were no occurrences of arm-biting or aggression in the analysis. All target behaviours were collapsed and coded as a single response. Inter-rater reliability was calculated for 29.6% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for challenging behaviour was 87.5%.

Table 5.4

Jacob. Total Subscale Scores from QABF Subscales.

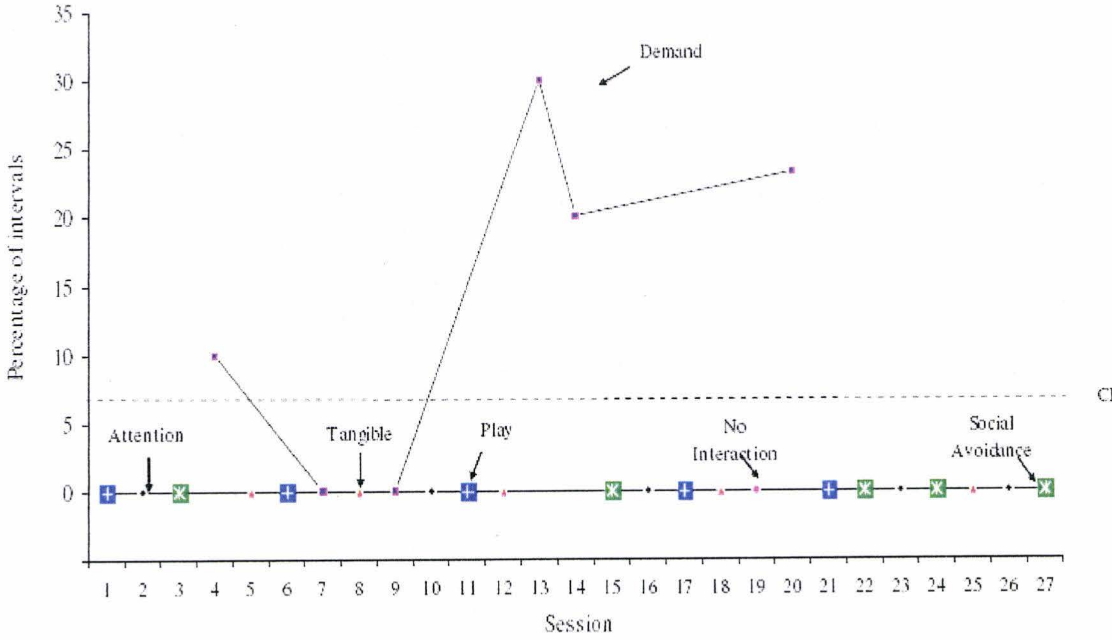
	Self-injurious behaviour	Aggression	Property Destruction
Jacob.			
Attention	3	0	N/A
Tangible	8	7	N/A
Demand	6	6	N/A
Physical discomfort	0	0	N/A
Automatic	4	5	N/A

All experimental sessions were conducted in a room in the family home. Academic tasks used in the demand condition were selected following initial assessment and involved a matching to sample task (e.g., matching coins and numbers). The tangible condition involved withholding access to toys/food items. Only a single no interaction condition was included in the analysis due to practical difficulties (Jacob would interact with family members on the other side of the door).

Figure 5.6 shows session by session data for the occurrence of challenging behaviour during the functional analysis. Challenging behaviours occurred exclusively in the demand condition of the functional analysis (M = 13.9; range = 0-30). There were no occurrences of challenging behaviour in any other condition of the functional analysis.

As at least 50% of scores for the demand condition lay above the CL the data met the criteria for escape-maintained challenging behaviour.

Figure 5.6. Jacob. Percentage of intervals with challenging behaviour during functional analysis.



Data for Jacob suggested that challenging behaviour may have been negatively reinforced by the contingent removal of aversive stimuli (e.g., demands). Interestingly there were no occurrences of arm-biting or aggression during any of the functional analysis conditions, despite this being reported by Jacob's mother as occurring several times a day. During informal observations by the lead researcher it appeared that both responses were occasioned by physical attacks by Jacob's brother. For ethical reasons this hypothesis could not be experimentally tested during the functional analysis.

Luke.

Luke was fifteen years old and had a diagnosis of FXS. He was non-verbal and primarily communicated by leading people to objects which he wanted. Luke lived in the family home and attended a school for children with severe learning disabilities. The QABF was completed as part of study one by Luke's mum, which indicated that Luke primarily displayed challenging behaviours maintained by escape and access to tangibles. The total subscale scores for Luke are presented in Table 5.5.

Table 5.5

Luke. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
Luke.			
Attention	5	6	5
Tangible	9	13	1
Demand	11	13	3
Physical discomfort	0	0	0
Automatic	2	2	7

Luke's mother requested that the analysis be conducted at school. Target behaviours included self-injurious behaviour (hand-biting with vocalisation) aggression (pulling, hitting, and scratching at therapist), property destruction (throwing and ripping objects) and foot stamping. Target behaviours were collapsed and coded as a single response. Inter-rater reliability was calculated for 34.6% of sessions. Using the more stringent measure of reliability (R-Occ) the reliability for challenging behaviour was 90.9%.

It was reported that Luke was much more likely to display challenging behaviour in the classroom than when he was in other parts of the school (e.g., therapy room). A limited number of studies have previously noted differences in the function of challenging behaviour across different settings (e.g., Harding, Wacker, Berg, Barretto, & Ringdahl, 2005; Lang et al., 2008). As such the current experimental analysis aimed to determine a) the function served by challenging behaviour, b) any changes in the occurrence and function of challenging behaviour across settings (e.g., classroom and therapy room). An

ABAB reversal with an embedded multi-element design was used in order to examine each of these two questions.

The functional analysis was conducted over four visits. In visit one and three the functional analysis was conducted in the classroom, in visits two and four the functional analysis was conducted in the therapy room. The classroom had at least five other students in at any time and two teaching assistants as well as the teacher. In order to minimise disruption to other students the functional analysis was conducted in a partitioned part of the classroom. The therapy room was a relatively large room, furnished with large cushions. The lead researcher and research assistant were the only other people in the therapy room.

All experimental sessions were conducted in either the classroom or the therapy room. Academic tasks used in the demand condition reflected those typically used at school and involved asking Luke to point at pictures of classmates. The tangible condition involved withholding access to preferred crisps. All other experimental conditions were run in accordance with the protocol for the study and each lasted for five minutes. There were a minimum of three replications of each experimental condition across settings.

Figure 5.7. Luke. Mean percentage of intervals with challenging behaviour across settings

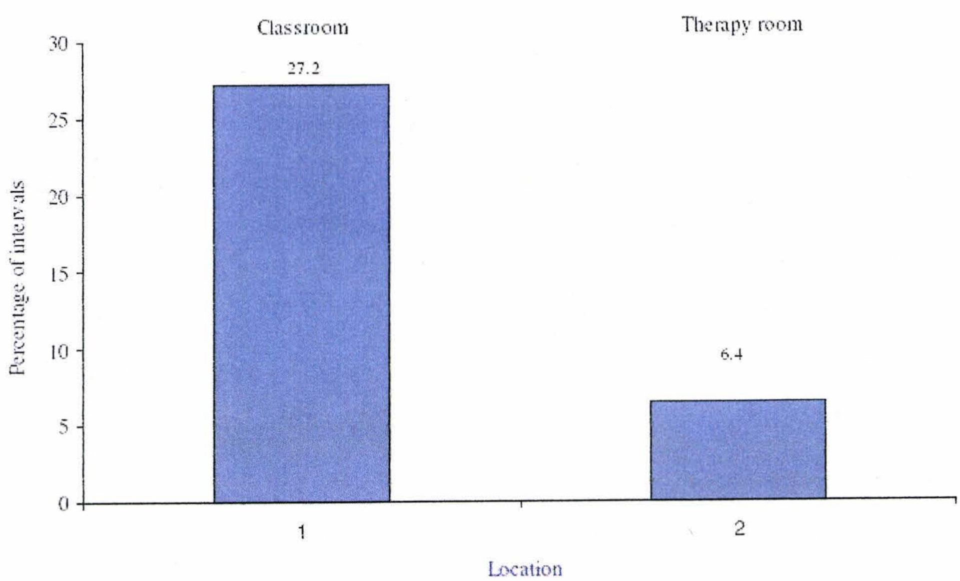


Figure 5.7 shows the overall occurrence of challenging behaviour in the classroom and therapy room. The overall mean percentage of intervals with challenging behaviour in the classroom (M = 27.2) was notably higher than when in the therapy room (M = 6.4).

Figure 5.8 shows session by session data for the occurrence of challenging behaviour during the functional analysis in both the classroom and therapy room.

Figure 5.8. Luke. Percentage of intervals with challenging behaviour during functional analysis in the classroom and therapy room.

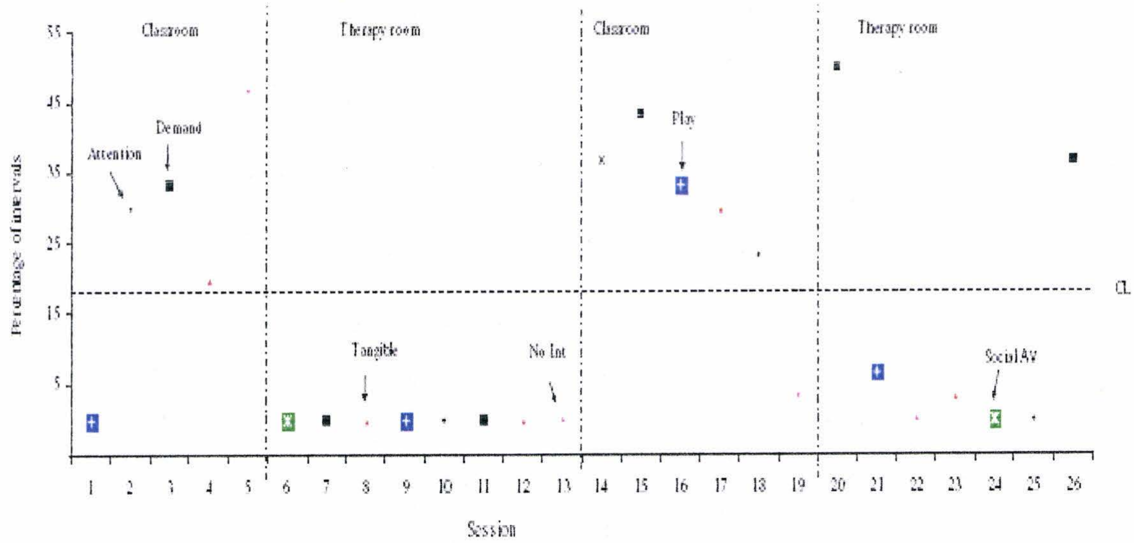
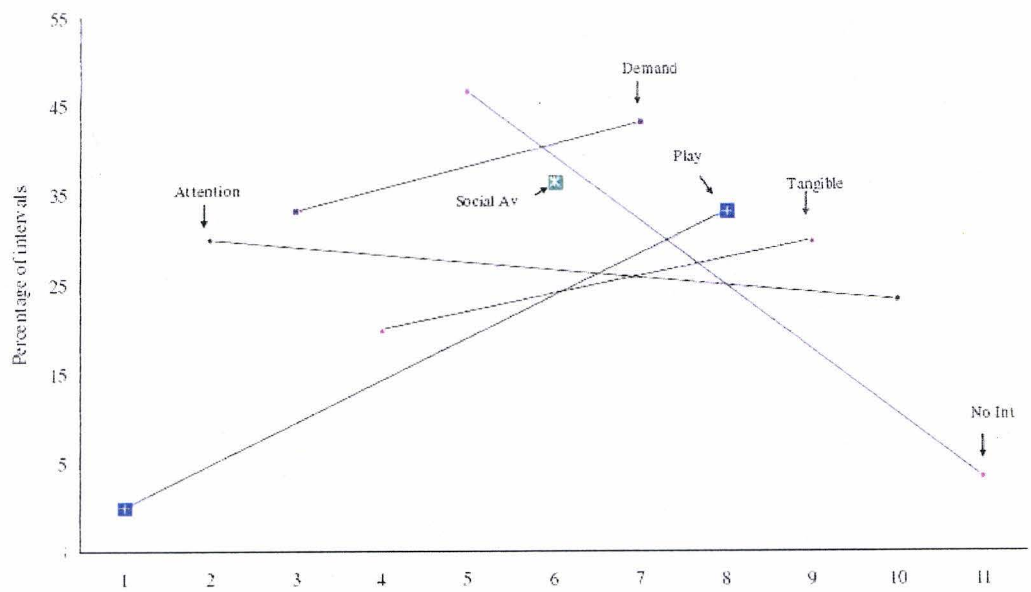


Figure 5.8 shows that with the exception of the demand condition, challenging behaviours were much less likely to occur in the therapy room than classroom. The functional analysis itself was generally characterised by high levels of variability, which were only in part attributable to the setting in which the analysis was conducted. There were high rates of challenging behaviour in all but two of the demand conditions (M = 27.2; range = 0 - 50), there were also relatively high levels of challenging behaviour in two of the five tangible conditions (M = 11.98; range = 0 - 30) and attention conditions (M=13.3; range = 0 - 30). Challenging behaviour occurred in two of the four no interaction (M = 9.97; range = 0 – 36.6) and play (M = 10; range = 0 – 33.3) conditions and one of the three social avoidance conditions (M = 12.2; range = 0 - 36.6).

As at least 50% of scores for the demand condition lay above the CL, the data met the criterion for escape-maintained challenging behaviour. Despite the observed variability no other condition met this criterion.

Figure 5.9 shows the results of those functional analysis conditions that were solely conducted in the classroom.

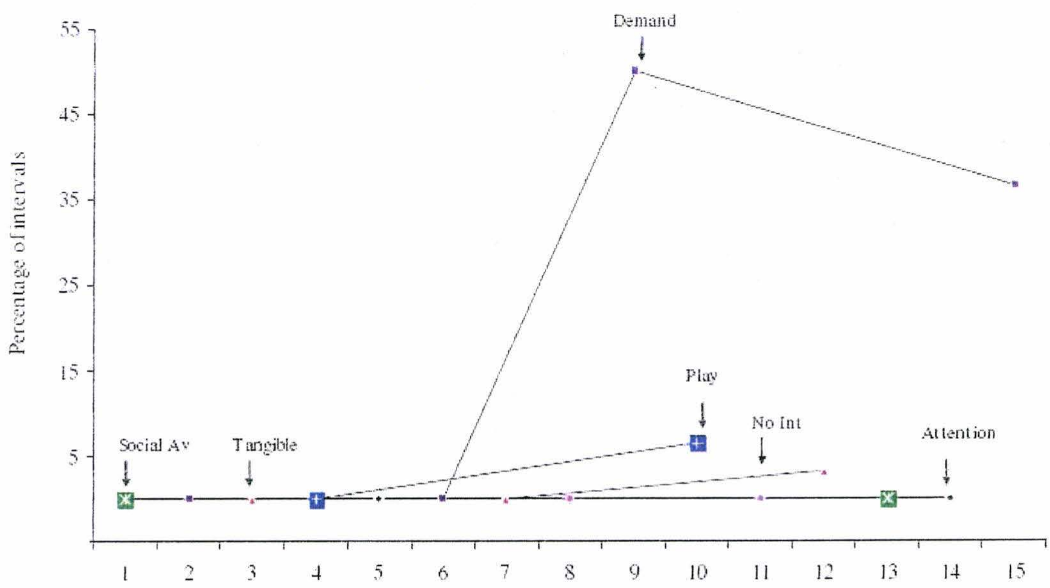
Figure 5.9. Luke. Percentage of intervals with challenging behaviour during classroom functional analysis.



There were heightened levels of challenging behaviour occurring in at least one of each experimental condition occurring in the classroom (*Demand*, M = 38.3; range = 33.3 – 43.3; *Social Avoidance*, M = 36.6: *Tangible*, M = 25.0; range = 20 - 30: *Play*, M = 16.7; range = 0 – 33.3: *No Interaction*, M = 25; range = 3.3 – 46.6: *Attention*, M = 26.7; range = 23.3 - 30). The absence of a clear 'control' condition in this setting makes a judgment about the function served by challenging behaviour difficult.

Figure 5.10 shows the results of those functional analysis conditions that were solely conducted in the therapy room. There were elevated levels of challenging behaviour in the demand condition (M= 21.7; range 0 - 50) in comparison to other experimental conditions (*Social Avoidance*, M = 0: *Tangible*, M = 1.1; range = 0 - 3.3: *Play*, M = 3.3; range = 0 - 6.6: *No Interaction*, M = 0: *Attention*, M = 0) in this particular setting.

Figure 5.10. Luke. Percentage of intervals with challenging behaviour during therapy room functional analysis.



In sum, whilst it appeared that Luke primarily displayed escape-maintained challenging behaviour, it was not possible to rule out alternative accounts of potential maintaining variables when in the classroom. In classroom settings there were high levels of variability across all experimental conditions. One explanation for this could be that challenging behaviour occurring in the classroom was automatically reinforced and not sensitive to environmental contingencies. Whilst certainly possible, it is unclear why challenging behaviours would cease to be automatically reinforcing when Luke was out of the classroom. This points to the influence of potential extraneous variables that may have influenced the occurrence of challenging behaviour in each setting. For example, when Luke was working in the partitioned 'functional analysis' area of the classroom he was prevented from accessing other parts of the classroom and thus potential reinforcers, such as food, classmates, teachers, books. Thus, it is possible that the restriction of movement and reinforcement functioned as an establishing operation for challenging behaviour (e.g., Fritz, DeLeon, & Lazarchick, 2004) and led to high levels of challenging behaviour occurring across all conditions of the functional analysis in these settings. When in the therapy room there were no partitions and thus with the removal of this particular EO, Luke's behaviour may have become sensitive to the more immediate contingencies of the functional analysis.

Theo.

Theo was ten years old and had a diagnosis of FXS. Theo was verbal and tended to communicate using single repetitive words. Theo attended a school for children with moderate learning disabilities and lived in the family home. The QABF was completed as part of study one by Theo's mum and indicated that Theo primarily displayed escape-maintained challenging behaviours. The total subscale scores for Theo are presented in Table 5.6.

Table 5.6

Theo. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
Theo.			
Attention	2	2	N/A
Tangible	0	0	N/A
Demand	9	9	N/A
Physical discomfort	0	0	N/A
Automatic	3	3	N/A

Target behaviours included destructive behaviours (ripping up paper), self-injury (finger-biting) and aggression (hitting others). There were no occurrences of finger-biting in the analysis. Target behaviours were collapsed and coded as a single response. Inter-rater reliability was calculated for 36% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability was 89.5%.

The functional analysis was conducted in a room in the family home. Following discussion with Theo's parents a matching-to-sample task involving numbers was selected for the demand condition of the functional analysis, and the tangible condition involved withholding access to preferred 'Pringles' crisps. As one of the target behaviours included ripping paper, a piece of paper was made available in each session for all conditions.

Figure 5.11. Theo. Percentage of intervals with challenging behaviour during functional analysis.

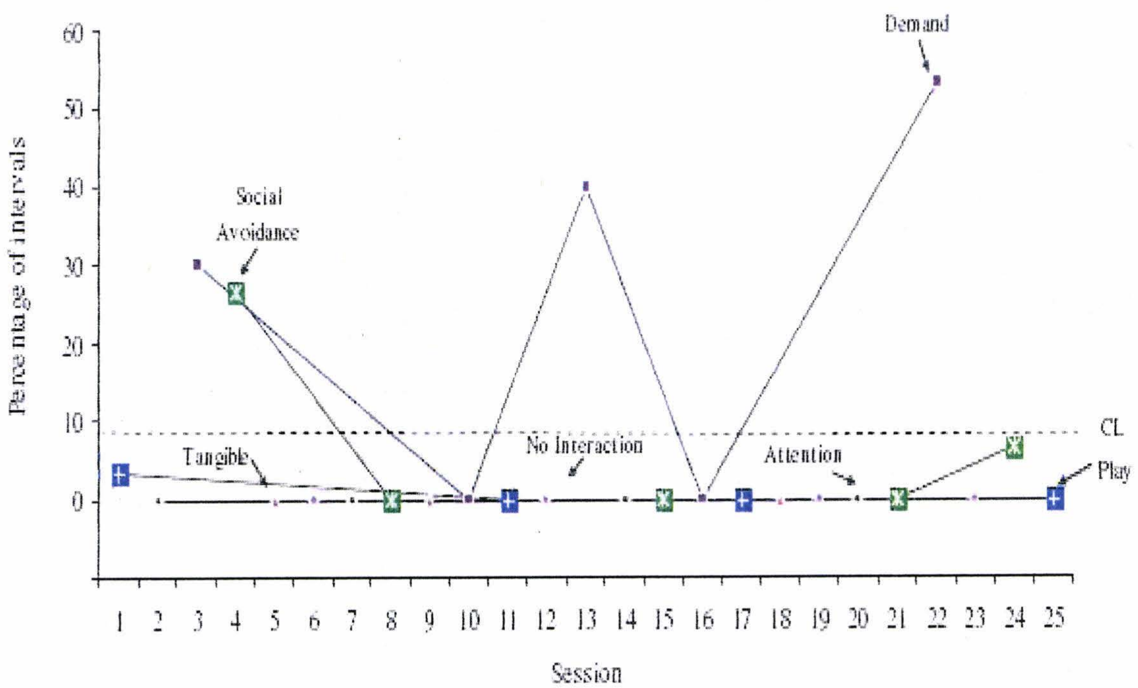


Figure 5.11 shows the results of the functional analysis for Theo. There were high rates of challenging behaviour in three of the five demand conditions (M = 24.7; range = 0 – 53.3). There were also elevated levels of challenging behaviour in two of the five social avoidance conditions (M = 6; range = 0 - 26.6). There were zero to low rates of

challenging behaviour in all other conditions (*Play*, M = .83; range = 0 - 3.3: *Attention*, M = 0: *Tangible*, M = 0: *No Interaction*, M = 0).

As at least 50% of scores for the demand condition lay above the CL, the data met the criterion for escape-maintained challenging behaviour.

It appears that Theo displayed challenging behaviour that was negatively reinforced by the removal of aversive stimuli, such as academic demands. Interestingly, during the functional analysis there were minimal occurrences of aggression and no occurrences of finger-biting. Theo's parents reported that he was especially likely to display such behaviours when asked to do something novel and when in situations that he found particularly 'stressful'. Anecdotally he was observed to repeatedly punch his father in the face when asked to work alone with the research team on the first visit to the family home. Unfortunately it was not possible to find a way to incorporate an analogue of this situation into the functional analysis itself.

John.

John was twelve years old and had a diagnosis of FXS. John was verbal and primarily communicated using repetitious sentences and one word utterances. He attended a school for children with moderate learning disabilities and lived in the family home. The QABF was completed as part of study one by John's mum and indicated that self-injury and aggressive behaviours were maintained by escape from demands and access to tangibles, whereas property destruction was automatically reinforced. The total subscale scores for John are presented in Table 5.7.

The target behaviours included for analysis were self-injurious behaviour (finger-biting), destructive behaviours (throwing objects, foot stamping) and aggression (hitting out at others). There were no occurrences of object throwing, foot-stamping or aggression in the analysis. Inter-rater reliability was calculated for finger-biting in 36.4% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability was 87.5%.

Table 5.7

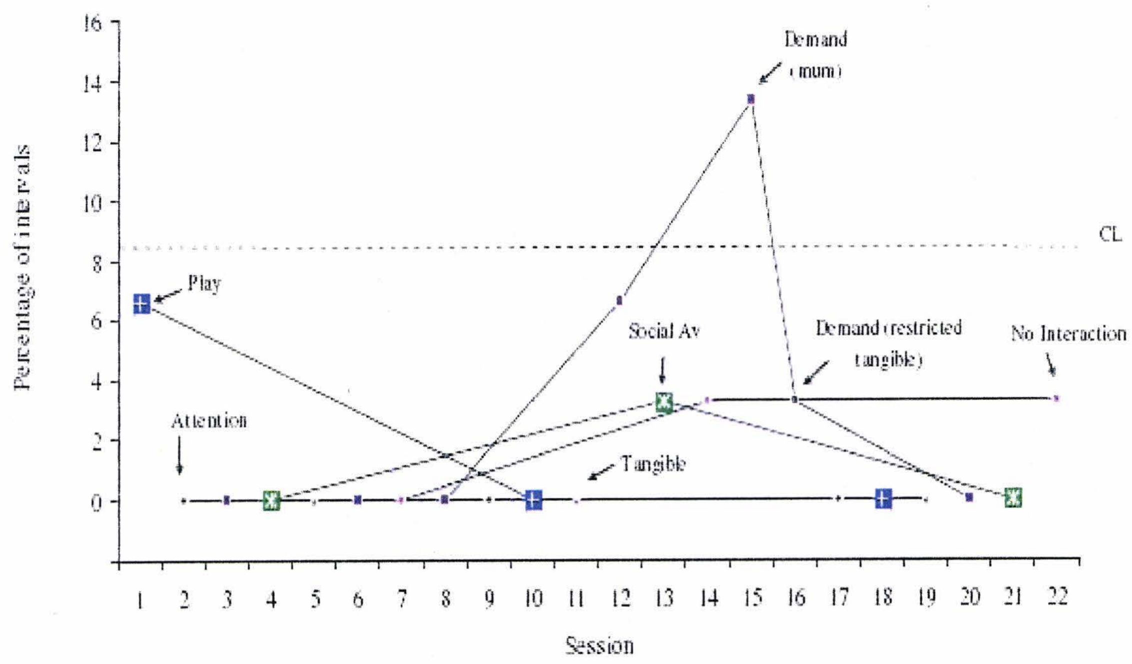
John. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
John.			
Attention	2	6	0
Tangible	12	10	2
Demand	12	13	2
Physical discomfort	0	0	0
Automatic	10	0	14

The functional analysis was conducted in a room in the family home. The tangible condition of the functional analysis involved withholding access to preferred items (television control, toy cars, crisps). A number of variants of the demand condition were included in the functional analysis in an attempt to provide a more externally valid analogue of those situations that evoked challenging behaviour in John's natural environment. One session involved John's mother presenting academic demands (without any explicit consequences programmed for any behaviour), another involved combining a standard demand condition with restricted access to the television and providing a break with access to the television contingent on challenging behaviour (cf., Call, Wacker, Ringdahl, & Boelter, 2005).

The results of the functional analysis, shown in Figure 5.12, suggest that finger-biting was primarily negatively reinforced and was occasioned by the onset of aversive stimuli (e.g., demands). There were elevated levels of challenging behaviour in three of the seven demand conditions (M = 3.31; range = 0 – 13.3) of the functional analysis. Finger biting only occurred in one play condition (M= 2.2; range = 0 - 6.6) and one social avoidance condition (M = 1.1; range = 0 – 3.3). Finger biting occurred in two no interaction conditions (M = 2.2; range = 0 – 3.3) of the functional analysis. There were zero rates of challenging behaviour in the tangible and attention conditions of the functional analysis.

Figure 5.12. John. Percentage of intervals with challenging behaviour during functional analysis.



As finger-biting occurred at a low-rate, the criteria suggested by Hagopian et al (1997) for low-rate behaviours was applied to ascribe function[30]. Using these criteria, there was differentiation in the demand condition of the analysis.

[30] "In cases in which most of the data points are low, the condition in which all or most of the higher rate behavior occurs is considered to be differentiated.... However, one of those high points must occur in the last half of the assessment." (see Hagopian et al., 1997, p. 325). Footnote continues overleaf.

It appears that John displayed finger-biting that was primarily negatively reinforced by the removal of aversive stimuli, such as academic demands. Finger-biting occurred at relatively low rates, however, and there were no occurrences of aggression or destructive behaviours. Indeed there were several demand conditions in which John displayed no challenging behaviour. This contrasted markedly with reports from John's mother and school teacher. Indeed, John was anecdotally observed by the lead researcher to engage in high rates of challenging behaviour (including throwing objects, and foot stamping) in natural settings at home both when asked to do something but also when unable to access preferred foods or activities, such as watching the television. However, there were no occurrences of challenging behaviour during any of the tangible conditions, despite restricting access to the same stimuli, and there were only low rates of finger-biting in demand conditions. It is unclear as to why this was the case. One possible account could be that discriminative stimuli that were present in the natural environment were not in situ during the functional analysis and as such John failed to contact the available contingencies. Another interpretation could be that some aspect of the functional analysis served to abolish the reinforcing value of some of the consequences that may have maintained challenging behaviour or alternatively may have established some of the other consequences of challenging behaviour as aversive.

Calum.

Calum was a fourteen year old boy with a diagnosis of FXS, who attended a school for children with moderate learning disabilities and lived in the family home. Calum was verbal and could communicate using relatively complex sentences. The QABF was completed as part of study one by Calum's mum and suggested that Calum's challenging

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For John, there were a total of 7 intervals in the demand condition in which finger biting occurred, in comparison to 2 intervals in both the play and no interaction conditions and 1 interval in the social avoidance condition.

behaviours were primarily escape-maintained. The total subscale scores for Calum are presented in Table 5.8.

Table 5.8

Calum. Total Subscale Scores from QABF Subscales.

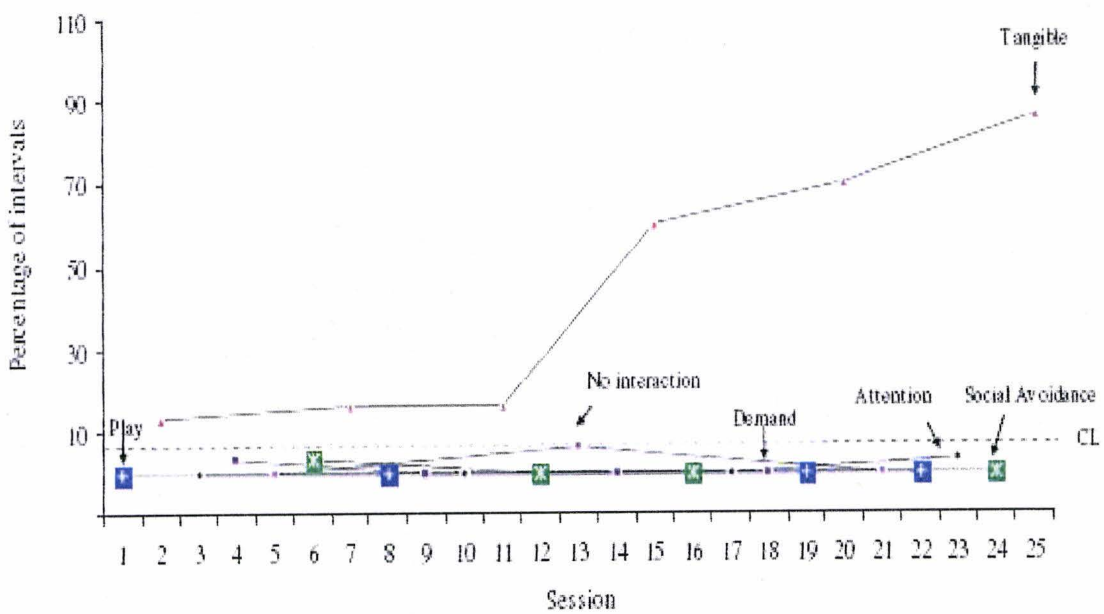
	Self-injurious behaviour	Aggression	Property Destruction
Calum.			
Attention	N/A	7	2
Tangible	N/A	6	6
Demand	N/A	11	11
Physical discomfort	N/A	2	2
Automatic	N/A	11	0

Target behaviours included self-injurious behaviours (banging head against furnishings), aggression (grabbing the lead researcher by the arm) and property destruction (throwing items, banging on surfaces with closed fist). There were no occurrences of self-injurious behaviours in the functional analysis. Target behaviours were collapsed and coded as a single response. Inter-rater reliability was calculated for 34.5% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability was 97.9%.

The functional analysis was conducted in a room in the family home. Following discussion with Calum's mother a matching-to-sample task was used in the demand condition and the tangible condition involved withholding access to preferred chocolate chip cookies. Due to the non-occurrence of self-injurious behaviour (which was the initial focus of the assessment) target behaviours were not consistently consequated until the fourth tangible condition.

The results of the functional analysis, shown in Figure 5.13, clearly show that challenging behaviours were positively reinforced by the provision of preferred tangible items. There were elevated rates of challenging behaviours occurring in each tangible condition (M = 43.85; range = 13.3 - 86.6). There were relatively low rates of challenging behaviour in the demand (M = .83; range = 0 – 3.3), social avoidance (M = .83; range = 0 - 3.3) and no interaction (M = 2.2; range = 0 - 6.6) conditions of the functional analysis. There were zero rates of challenging behaviour in the play and attention conditions of the functional analysis.

Figure 5.13. Calum. Percentage of intervals with challenging behaviour during functional analysis.



As at least 50% of scores for the tangible condition lay above the CL, the data met the criteria for tangible-maintained challenging behaviour.

It appears that Calum primarily displayed challenging behaviour that was maintained by the provision of tangible items (i.e., chocolate cookies). The results of this analysis are relatively clear; however it is unclear as to why Calum never engaged in any self-injurious behaviours, which had been reported as frequently occurring in natural settings.

Richard.

Richard was an eleven year old boy with a diagnosis of FXS, who attended a school for children with moderate learning disabilities and lived in the family home. Richard was verbal and would communicate using relatively complex sentences. The QABF was completed as part of study one by Richard's mum and self-injurious behaviours appeared to be multiply controlled. No clear function could be identified for aggressive behaviours using the QABF. The total subscale scores for Richard are presented in Table 5.9.

Table 5.9

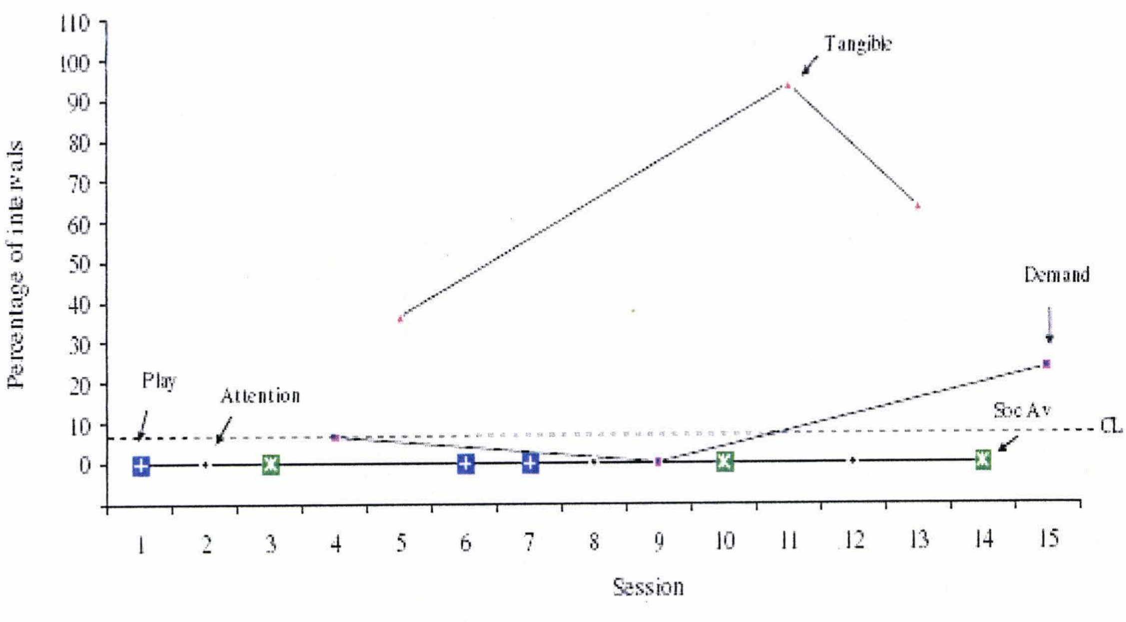
Richard. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
Richard.			
Attention	7	0	N/A
Tangible	8	2	N/A
Demand	9	3	N/A
Physical discomfort	10	1	N/A
Automatic	3	0	N/A

Target behaviours included aggression (kicking, hitting, ear pulling, pushing, spraying water, and throwing items at the therapist), self-injury (biting his little finger whilst sticking the index finger of his other hand up at the therapist), and property destruction (throwing items, ripping materials, spitting water). Target behaviours were collapsed and coded as a single response. Inter-rater reliability was calculated for 46.6% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability was 96%.

The functional analysis was conducted in the living room of the family home. Following initial assessment a matching-to-sample task was used in the demand condition and the tangible condition involved withholding access to preferred items (such as building blocks and a portable games console). A no interaction condition could not be included as the layout of the room meant that it was difficult to prevent Richard from leaving the assessment area to interact with other members of the family.

Figure 5.14. Richard. Percentage of intervals with challenging behaviour during functional analysis.



The results of the functional analysis, shown in Figure 5.14, clearly show that challenging behaviours were positively reinforced by the provision of preferred tangible items. There were elevated rates of challenging behaviours occurring in each of the three tangible conditions (M = 64.4; range = 36.6 - 93.3). There were slightly elevated rates of challenging behaviour in two of the three demand conditions (M = 9.96.; range = 0 –

23.3)[31]. There were zero occurrences of challenging behaviour in the play, attention, and social avoidance conditions of the functional analysis.

As at least 50% of scores for the tangible condition lay above the CL, the data met the criteria for tangible-maintained challenging behaviour.

It appears that Richard primarily displayed challenging behaviour that was maintained by the provision of tangible items.

Participants with FXS. Within-Group Analysis

A within-group analysis of the pattern of self-injury displayed by each participant is also possible using summary data. Figures 5.15-5.23 provide summary data for each participant in the FXS group. The numerical data in each figure indicates the overall mean percentage of intervals of challenging behaviour and the overall standard deviation. The data for each experimental condition are also presented graphically to present the number of standard deviations each condition mean is from the overall mean (i.e., the Z score for each experimental condition). As such, Figures 5.15-5.23 provide a summary of both the absolute level and relative variability of each participant's challenging behaviour (Iwata et al., 1982/1994).

In what follows, particular emphasis is given to the analysis of those topographies of challenging behaviour that appear to be socially maintained. All topographies that appeared to be automatically reinforced were excluded from the current analysis. A number of apparent patterns in the data are discussed.

Figures 5.15-5.18 present summary data for four participants with FXS, for whom at least one response class of challenging behaviour was positively reinforced by the contingent provision of tangible items. Figures 5.19-5.23 present summary data for five participants

[31]Although unclear in Figure 5.14, the percentage of intervals with challenging behaviour in session 4 (demand condition) is at 6.6% of intervals and falls below the CL of 8.3% of intervals.

who displayed at least one response class of challenging behaviour that appeared to be negatively reinforced by the removal of either social attention or academic demands.

Figure 5.15. Abe. No. of standard deviations each condition mean was from the overall mean for other challenging behaviours.

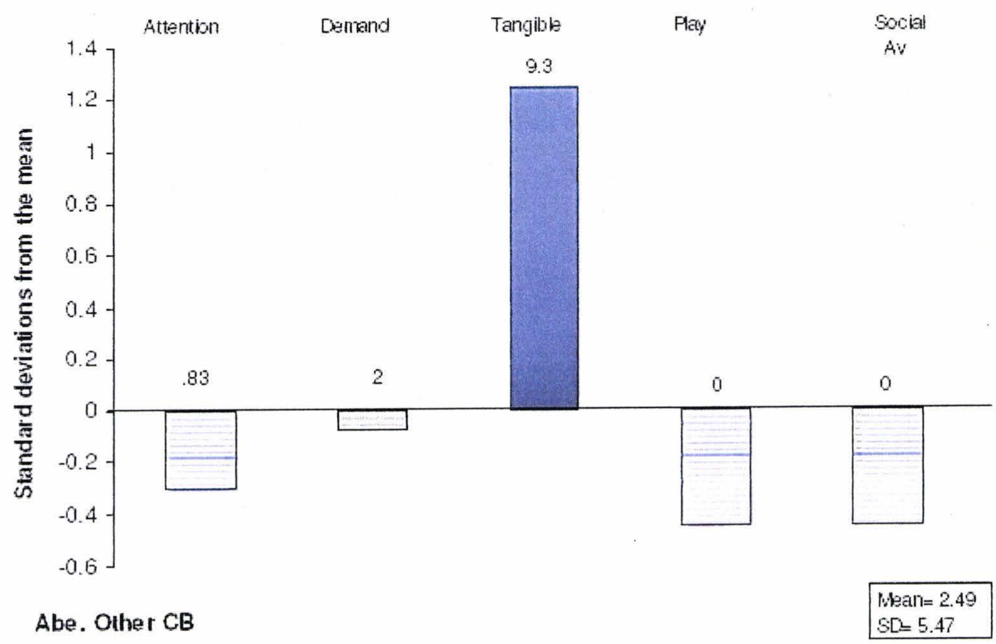


Figure 5.16. Calum. No. of standard deviations each condition mean was from the overall mean for challenging behaviour.

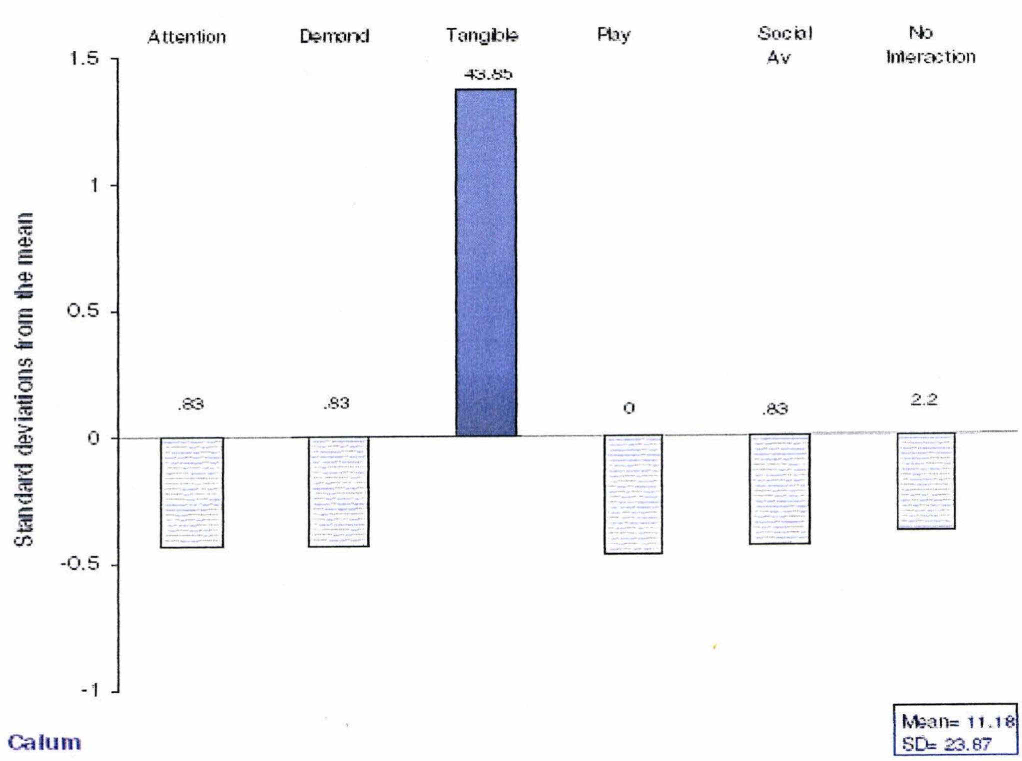


Figure 5.17. Greg. No. of standard deviations each condition mean was from the overall mean for other challenging behaviours.

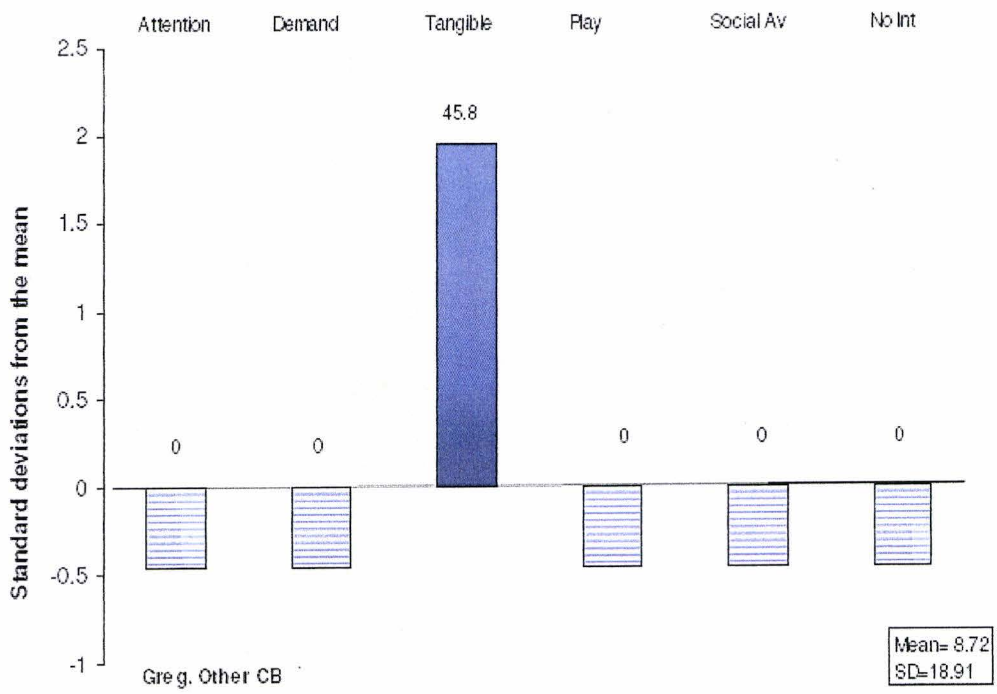
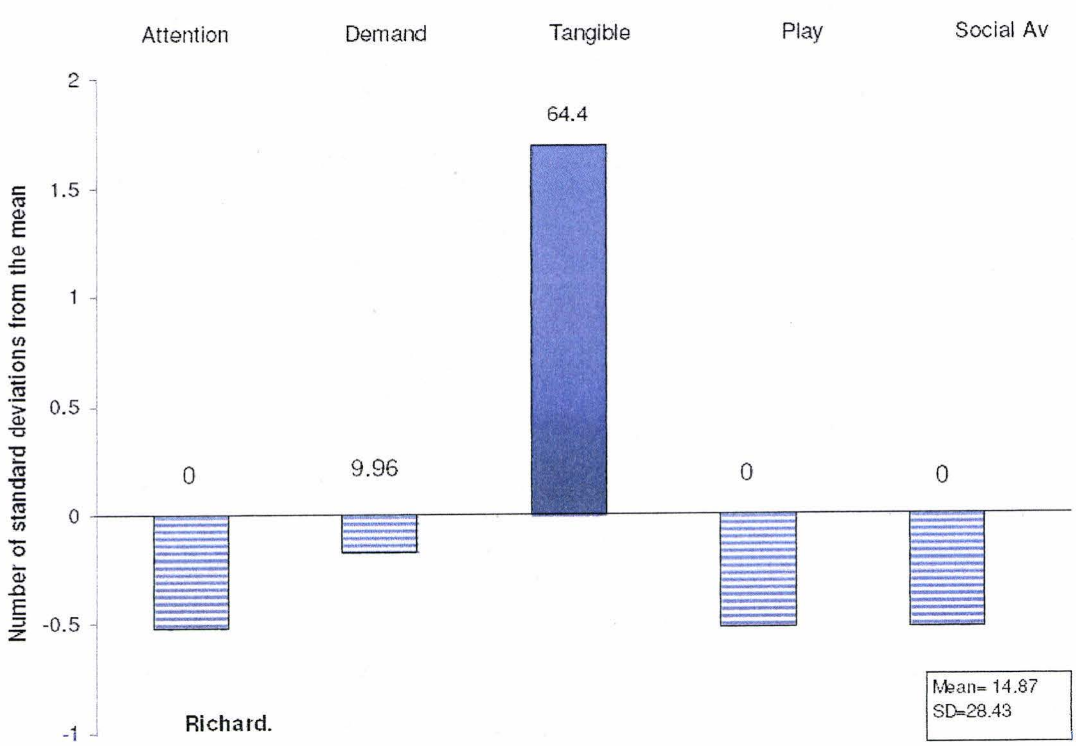


Figure 5.18. Richard. No. of standard deviations each condition mean was from the overall mean for challenging behaviour.



Figures 5.15-5.18 shows the number of standard deviations each condition mean was from the overall mean for Abe, Calum, Greg and Richard's challenging behaviour. It appears that each of these participants displayed at least one topography of challenging behaviour that was positively reinforced by the provision of tangible items.

For Abe, as shown in Figure 5.15, the mean occurrence of challenging behaviours (excluding finger-biting) in the tangible condition was 9.3 which was 1.24 standard deviations above the overall mean of 2.49. For Calum, as shown in Figure 5.16, the mean occurrence of challenging behaviours in the tangible condition was 43.85 which was 1.37 standard deviations above the overall mean of 11.18. For Greg, the mean occurrence of other challenging behaviours in the tangible condition was 45.8, which was 1.96 standard deviations above the overall mean of 8.72, as shown in Figure 5.17. For Richard, as shown in Figure 5.18 the mean occurrence of challenging behaviour in the tangible condition was 64.4 which was 1.7 standard deviations above the overall mean of 14.87. These data support the interpretation that at least one response class for each of these four participants was maintained by access to tangible items.

Figure 5.19. Jacob. No. of standard deviations each condition mean was from the overall mean for challenging behaviour.

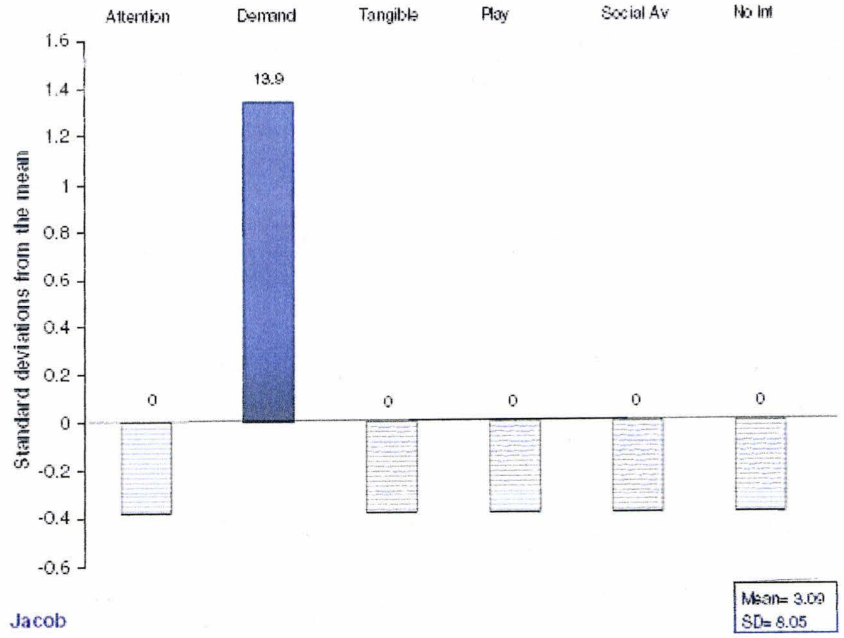


Figure 5.20. Luke. No. of standard deviations each condition mean was from the overall mean for challenging behaviour.

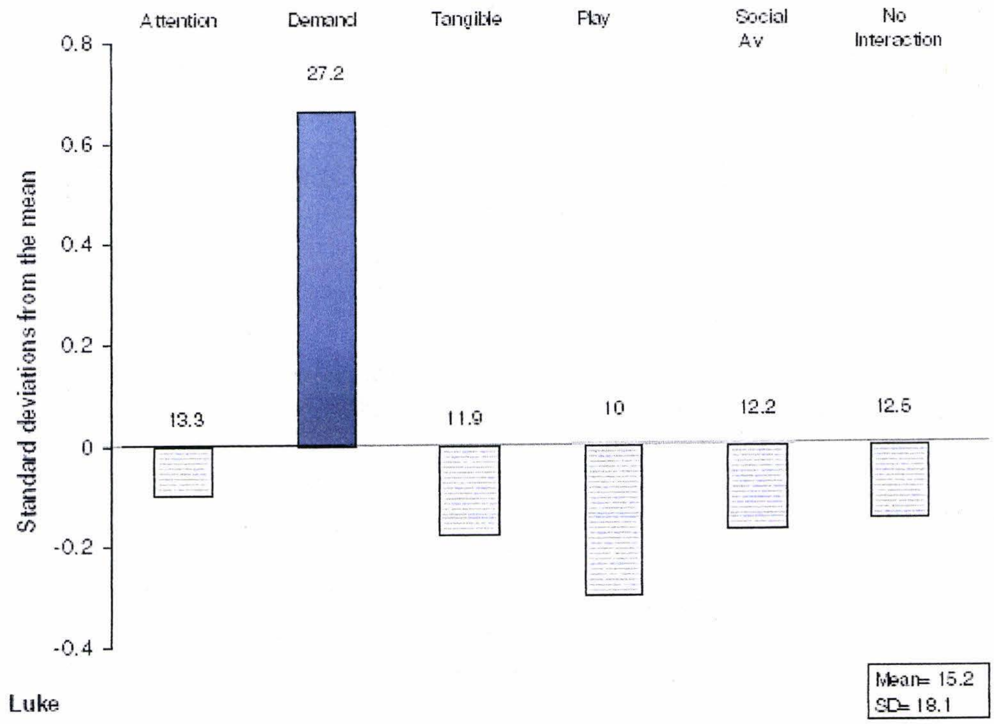


Figure 5.21. Theo. No. of standard deviations each condition mean was from the overall mean for challenging behaviour.

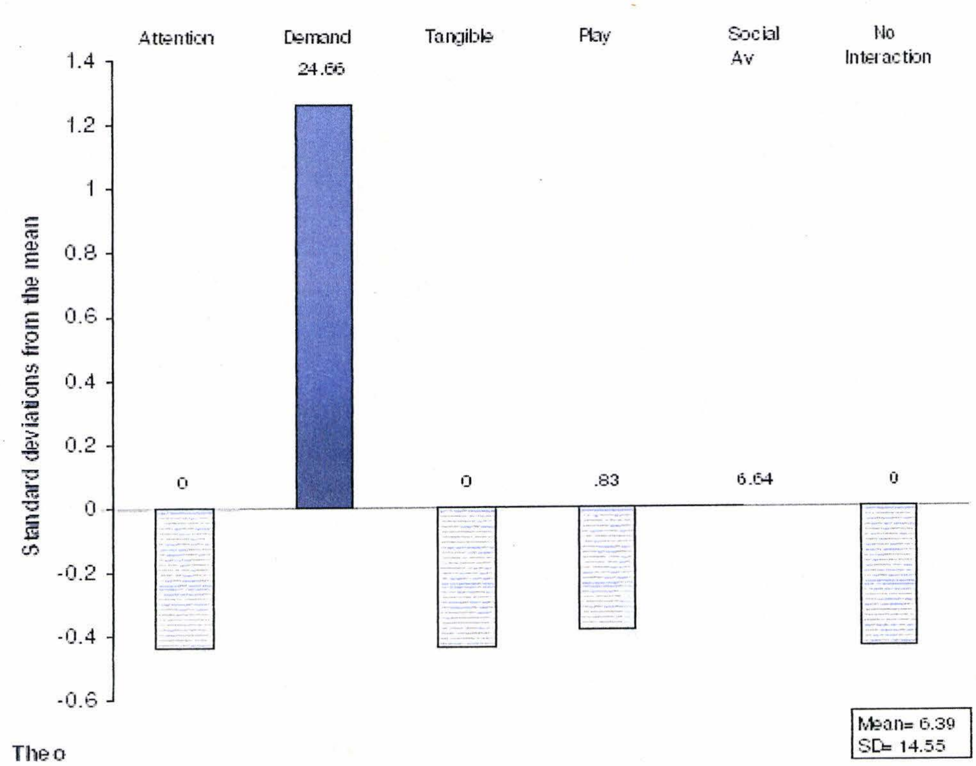


Figure 5.22. John. No. of standard deviations each condition mean was from the overall mean.

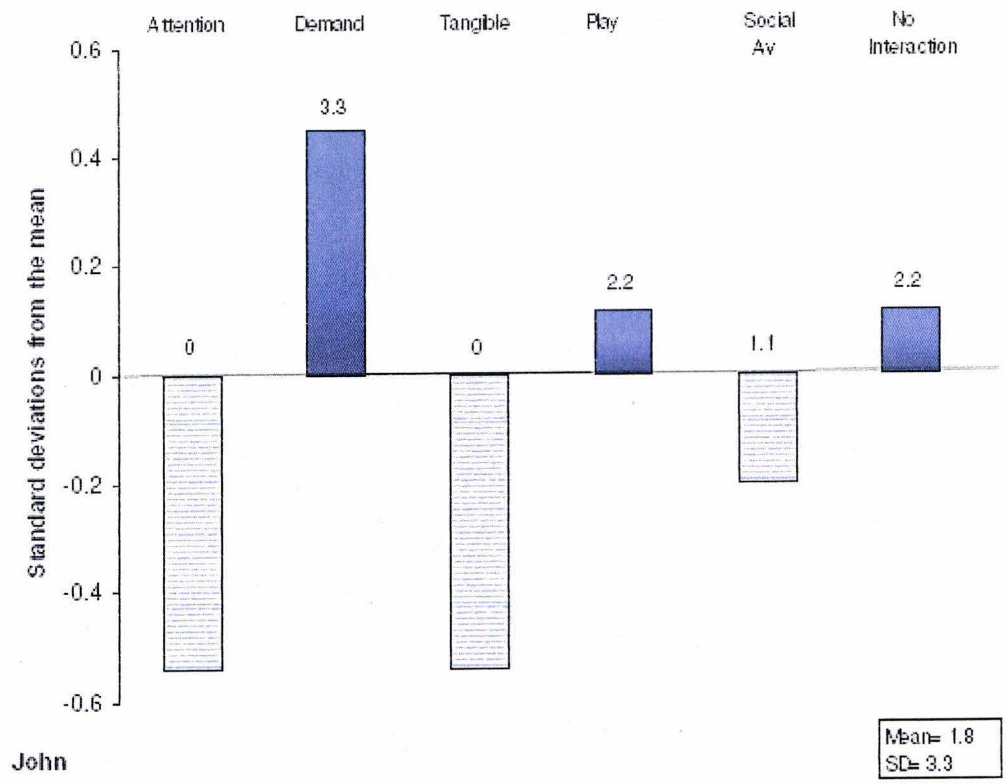
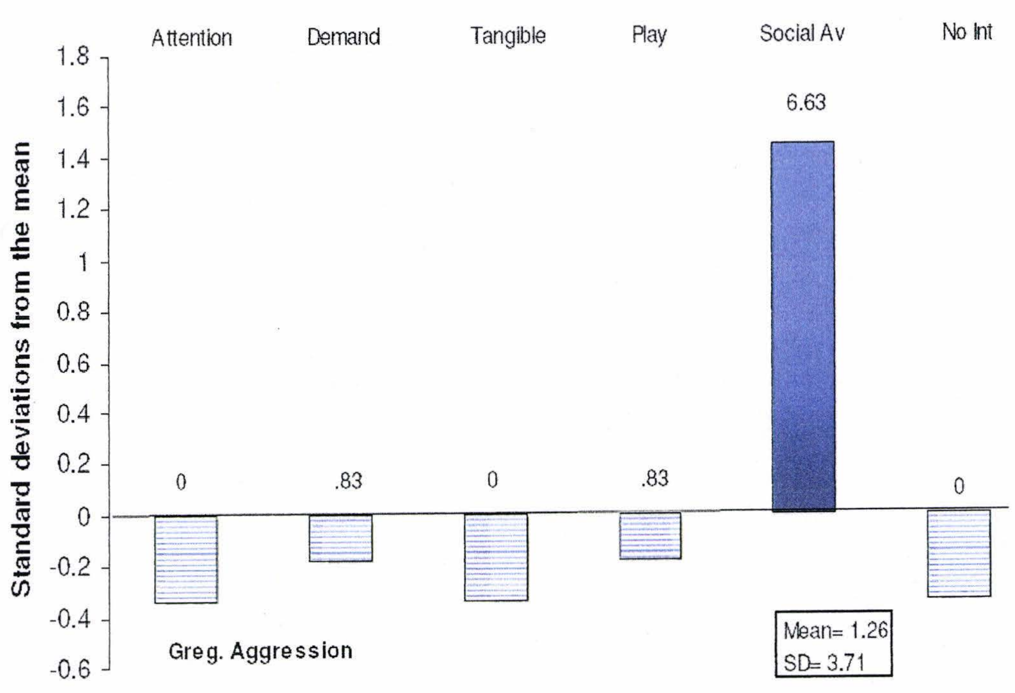


Figure 5.23. Greg. No. of standard deviations each condition mean was from the overall mean for aggression.



Figures 5.19-5.23 present summary data for those participants (Jacob, Luke, Theo, John, and Greg) who displayed at least one topography of challenging behaviour that appeared to be negatively reinforced by the removal of aversive stimuli, such as attention or demands.

For Jacob, as shown in Figure 5.19, the mean occurrence of challenging behaviour in the demand condition was 13.9, which was 1.34 standard deviations above the overall mean of 3.09. For Luke, as shown in Figure 5.20, the mean occurrence of challenging behaviour in the demand condition was 27.2, which was .7 standard deviations above the overall mean of 15.2. Figure 5.21 shows that the mean occurrence of challenging behaviour in the demand condition for Theo was 24.66, which was 1.26 standard deviations above the overall mean of 6.39. Figure 5.22 shows that the mean occurrence of challenging behaviour in the demand condition for John was 3.3, which was .45 standard deviations above the overall mean of 1.8. Finally for Greg, as shown in Figure 5.23, the mean occurrence of aggression in the social avoidance condition was 6.63, which was 1.45 standard deviations above the overall mean of 1.26. Overall this supports the interpretation that challenging behaviour displayed by Jacob, Luke, Theo, John and Greg were at least in part negatively reinforced by the removal of either attention or demands.

No participant with fragile X syndrome displayed any challenging behaviours that were maintained by social attention. Figures 5.18-5.23 show that for all those participants with a socially-influenced response class of challenging behaviour, the mean occurrence of challenging behaviours in the attention condition fell beneath the overall mean occurrence of challenging behaviour across conditions.

In sum, analysis of within-group variation for participants with FXS suggested three patterns of results. First a relatively high proportion of participants (4/8) displayed at least one response class of challenging behaviour that appeared to be tangible-maintained. Second, a relatively high proportion of participants (5/8) displayed at least one response class of challenging behaviour that appeared to be negatively-reinforced by the removal of demands or social attention. Finally no participants with FXS displayed challenging behaviours that were found to be positively reinforced by the provision of social attention. Indeed for all participants the mean attention condition scores for all socially influenced response classes of challenging behaviour fell below the overall mean.

Participants with SMS. Individual Functional Analyses

A total of six individuals with a diagnosis of SMS took part in study two. All participants' parents had taken part in study one, aside from Shelley who happened to attend the same school as another participant in the study. Table 5.10 provides some basic demographic information for each participant.

Table 5.10

Characteristics of Participants with SMS.

Participant	Age*	Total ABC	Age equivalent (Vineland Sub-domains)		
			Communication	Daily Living	Socialization
Sean	7yrs 9mths	68	1yr 11mths	3yrs 3mths	4yrs 8mths
Matt	9yrs 11mths	60	6yrs 8mths	3yrs 3mths	5yrs 5mths
Angus	11yrs 9mths	48	5yrs 1mth	4yrs 9mths	5yrs 5mths
Patrick	11 yrs 6mths	68	7yrs 8mths	4 yrs 2mths	4yrs 10mths
Shelley**	9yrs				
Katie***	11 yrs 7mths		6yrs 0mths	4yrs 6mths	5yrs 5mths

*Age at beginning of study two

**Due to time constraints Shelley's mother opted not to take part in study one.

***Data missing for Total ABC

Categorical data for five of the six children with SMS (excluding Shelley) were compared against the remainder of the SMS group from study one across all QABF subscales for each behavioural topography to ensure that the sub-group were representative of other participants with SMS included in the previous study. Using a significance level of $p = .05$, a series of chi-square analyses revealed no significant differences between the participants for any scale except the automatic subscale for self-injurious behaviour (with the subset of children taking part in study two being more likely to display automatically reinforced behaviour). Given that the primary emphasis of study two was on socially-mediated forms of reinforcement the participants with SMS taking part in study two were deemed to be sufficiently representative of the underlying group.

Sean.

Sean was a young boy aged seven years old with a diagnosis of SMS. Sean was non-verbal but was able to communicate using makaton signing. Sean attended a school for children with moderate learning disabilities and lived in the family home. The QABF was completed as part of study one by Sean's mum, and this indicated that self injurious behaviours appeared to be maintained by access to tangible-items and attention, aggressive behaviours appeared to be multiply socially controlled (tangible-, attention-, and escape-maintained). The total subscale scores for Sean are presented in Table 5.11.

Target behaviours included self-injury (finger and hand biting, head banging and head hitting), aggression (hitting and pushing), property destruction (kicking and hitting objects with an open palm, ripping, pulling at furnishings and throwing objects), non-compliance (dropping to the floor or turning away from researcher whilst refusing to go to work area) and crying (vocalisation with hands covering eyes). Target behaviours were collapsed into self-biting and other challenging behaviours. Inter-rater reliability was calculated for 44.4% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for self-biting was 83.3% and for other challenging behaviours was 85.1%.

Table 5.11

Sean. Total Subscale Scores from QABF Subscales.

		Self-injurious behaviour	Aggression	Property Destruction
Sean.				
	Attention	9	10	N/A
	Tangible	10	14	N/A
	Demand	4	15	N/A
	Physical discomfort	2	11	N/A
	Automatic	1	4	N/A

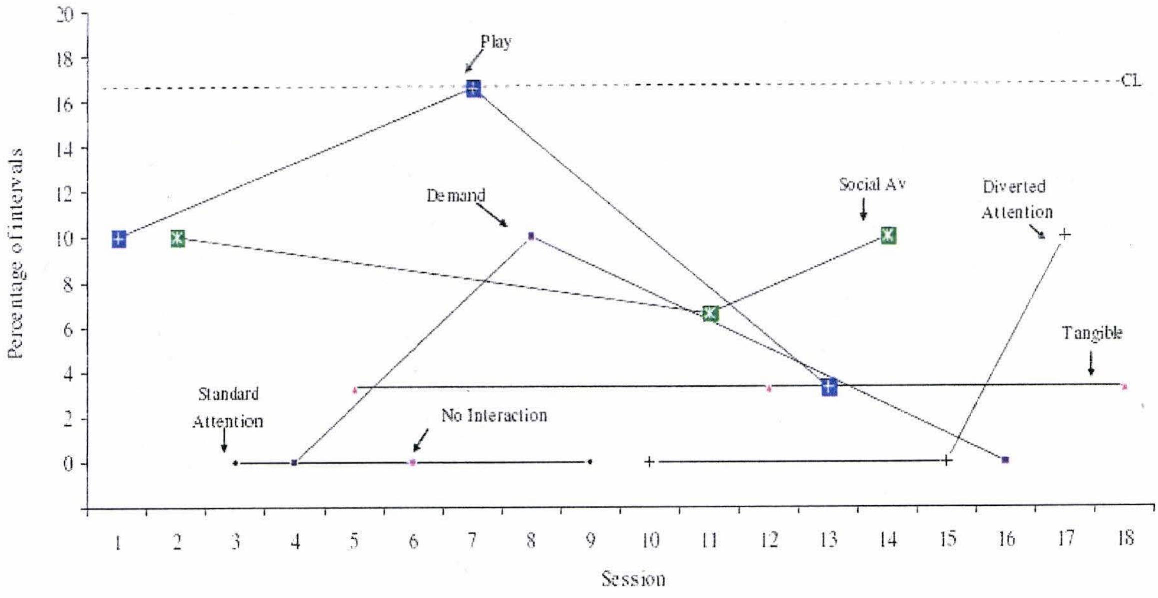
The functional analysis was conducted in a separate part of the family home that had been specifically designed for Sean. Following discussion with Sean's mother the standard attention condition was replaced with a diverted attention condition in which the lead researcher spoke continuously with the research assistant and provided attention to Sean contingent on challenging behaviour (cf., O'Reilly, Lancioni, King, Lally, & Dhomhnaill, 2000). Following initial observations by the research team the demand condition involved a matching-to-sample task that involved matching numbers and a spinning top toy was withheld in the tangible condition of the functional analysis. Due to practical difficulties (Sean would lock himself in the bathroom, where he could not be observed) only a single session of the no interaction condition was run.

Over the course of the functional analysis the severity of Sean's challenging behaviour increased notably and it became extremely difficult to keep Sean in the room for a whole session of the functional analysis (only two sessions could be completed in the final visit). Due to the practical difficulties this posed it was decided that the functional analysis should be terminated earlier than would otherwise have been the case.

Results of the functional analysis suggested that challenging behaviour formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.24 shows session by session data for the occurrence of finger-biting. There were high levels of self-biting in the play (M = 10; range = 3.3 – 16.6) and social avoidance (M = 8.9; range = 6.6 - 10) conditions of the functional analysis. There were also slightly elevated rates of this response in the tangible (M = 3.3; range = 3.3) condition and in a single demand (M = 3.3; range = 0 - 10) and single diverted attention condition (M = 3.3; range = 0 – 10) condition. There were no occurrences of self-biting in the no interaction condition or standard attention condition.

Figure 5.24. Sean. Percentage of intervals with self biting during functional analysis.



Due to the high level of challenging behaviour occurring in the play condition there was no differentiation between experimental conditions (Hagopian et al., 1997). This suggested that further analysis was merited in order to explore a potential automatically reinforced function.

Due to the unclear pattern of responding for self-biting an analysis of within-session data was conducted to test the hypothesis that self-biting was occasioned by the presence of specific stimuli. It appeared that Sean displayed self-biting more frequently in sessions in which he interacted with certain preferred toys that shared similar characteristics (i.e., produced movement and sound), such as a spinning toy, balloons and a Mr Face™ toy. The percentage of 10s intervals in which Sean interacted with these stimuli is shown in Figure 5.25, along with the percentage of intervals in which self-biting occurred. Self-biting appears to have been more likely to occur in sessions in which Sean had access and interacted with these toys than in sessions in which there was no toy interaction.

Figure 5.25. Sean. Percentage of intervals with toy interaction and finger biting across all conditions.

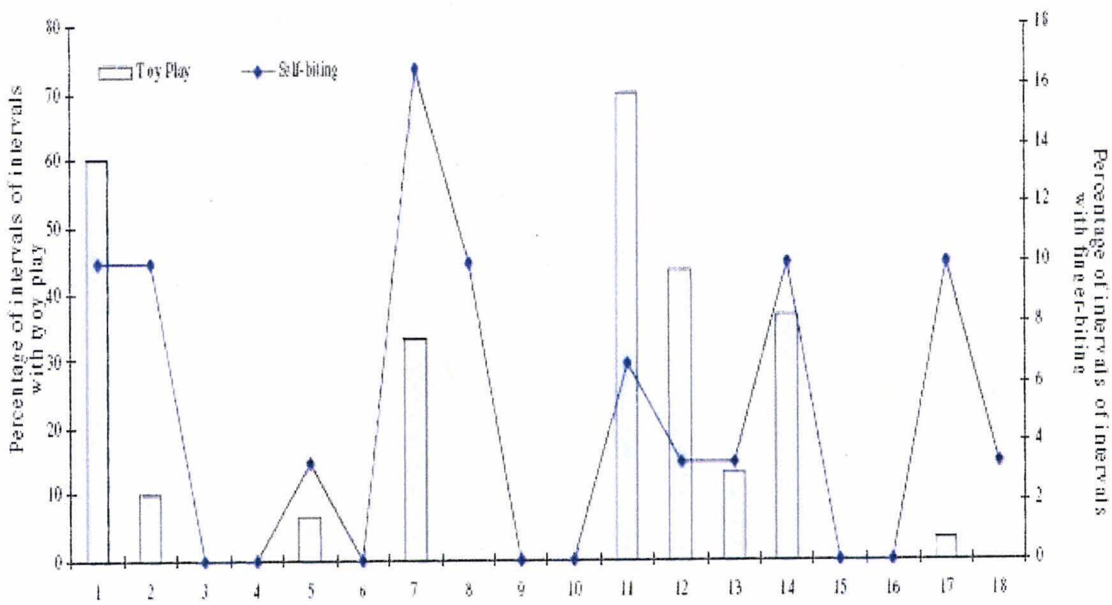


Figure 5.26. Sean. Percentage of intervals with other challenging behaviours during functional analysis.

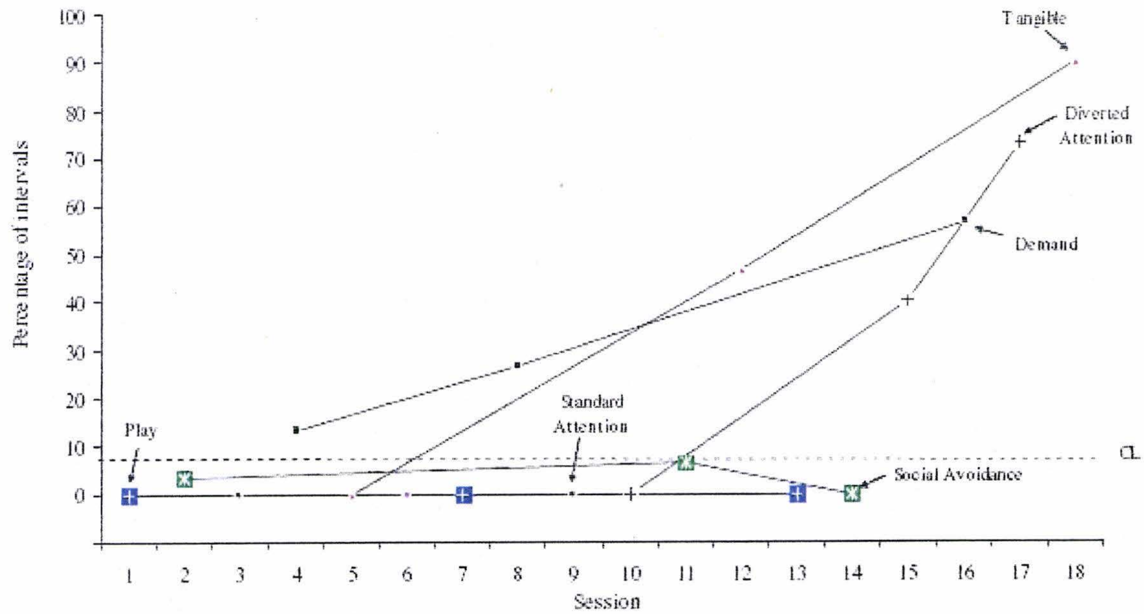


Figure 5.26 shows session by session data for the occurrence of all other topographies of challenging behaviour, excluding self-biting, displayed by Sean. There were heightened levels of challenging behaviour in the tangible (M = 45.5; range = 0 - 90), demand (M = 32.2; range = 0 - 56.6) and diverted attention (M = 37.6; range = 0 – 73.3) conditions of the functional analysis. There were low levels of challenging behaviour in the social avoidance (M = 3.3; range = 0 - 6.6) condition and zero rates in the play, no interaction and standard attention conditions of the functional analysis.

As at least 50% of scores for the each of the diverted attention, tangible and demand condition lay above the CL the data met the criteria for multiply-socially controlled challenging behaviour[32].

In sum, data for Sean suggested that self-biting was automatically reinforced and appeared to be especially likely to be occasioned by the presence as opposed to absence of particular stimuli, a pattern of responding that corresponds to that reported in other studies

[32] "In cases in which more than one condition meets criteria for differentiation, score the analysis as multiply maintained." See Hagopian et al (1997, p. 325).

(Friman, 2000; Van Camp et al., 2000). Other topographies of challenging behaviour appeared to form an alternative response class and were multiply socially controlled, occurring at differentially high levels in demand, attention and tangible conditions of the functional analysis. It is interesting to note that there were notable increases in the frequency, episodic severity (cf., LaVigna & Willis, 2005) and number of topographies of Sean's challenging behaviour as the functional analysis progressed. In addition, Sean would display challenging behaviours when these same conditions were being prepared but before the programmed antecedents were delivered. For example, when academic materials were pulled out of a bag, or certain toys were cleared away. It may have been that the preparation for certain conditions functioned as a reflexive CEO (or warning stimulus) whose onset established its own offset as an effective type of reinforcement (see Carr, Newsom, & Binkoff, 1980) and thereby evoked challenging behaviour.

Matt.

Matt was a boy aged nine years old with a diagnosis of SMS. Matt had good receptive and expressive communication skills, and was generally able to make his needs known. Matt attended a school for children with moderate learning disabilities and lived in the family home. The QABF was completed as part of study one by Matt's mum, and this indicated that challenging behaviours appeared to be multiply socially controlled (attention-, tangible-, and escape-maintained). Matt's challenging behaviours were also reported to be influenced by his sleep cycle and he scored highly on the physical discomfort related-subscale of the QABF. The total subscale scores for Matt are presented in Table 5.12.

Target behaviours included self-injury (banging body against objects), aggression (biting, hitting, spitting, kicking, scratching, pinching, head butting, pushing), property destruction (hitting objects with open palm, throwing large objects, ripping work materials, jumping), and non-compliance (crawling underneath table). Target behaviours were collapsed into object banging and other challenging behaviours. Inter-rater reliability was calculated for 48% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for object banging was 75.9% and for other challenging behaviours was 87.5%.

Table 5.12

Matt. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
Matt			
Attention	12	12	12
Tangible	9	9	9
Demand	10	10	10
Physical discomfort	15	15	15
Automatic	6	0	2

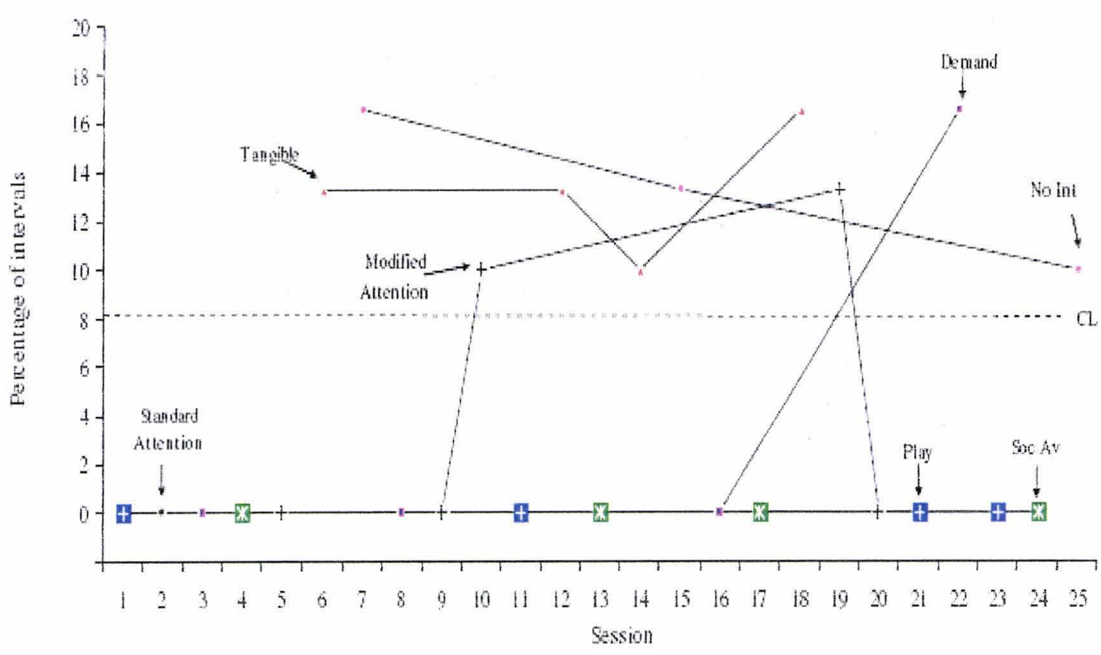
The functional analysis was conducted at school in a small room, which was used as Matt's classroom. The room was attached to a padded time-out room which remained locked throughout the majority of experimental sessions.

Following discussion with Matt's teaching assistant the standard attention condition was replaced with a modified attention condition in which the lead researcher asked Matt to do 'some work', whilst taking on the appearance of reading a paper. Matt was free to leave the work station at any point and no additional demands were given during the session. Attention was provided contingent on challenging behaviour whilst all other behaviours were ignored. Similar modified attention conditions have been used in other functional analysis studies (cf., Call et al., 2005) and it was felt that this arrangement better reflected the contingencies that governed Matt's behaviour in the natural environment. The demand condition used a series of arithmetic worksheets prepared by Matt's teacher. The tangible condition involved withholding access to balloons or a spinning top toy. All other conditions were run in accordance with the study protocol.

Results of the functional analysis suggested that challenging behaviour formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.27 shows session by session data for the occurrence of object banging. There were high levels of object banging in all tangible (M = 13.3; range = 10 – 16.6) and no interaction (M = 13.3; range = 10 - 16.6) conditions of the functional analysis. There were also slightly elevated rates of this response in two of the modified attention conditions (M = 4.7; range = 0 - 13.3) and in a single demand condition (M = 4.2; range = 0 – 16.6). There were no occurrences of object-banging in the play, social avoidance condition or standard attention condition.

Figure 5.27. Matt. Percentage of intervals with object banging behaviours during functional analysis.



The Hagopian et al (1997) criteria for the analysis of multiply-controlled challenging behaviour was used to analyse the data[33]. As at least 50% of scores for the tangible and no interaction conditions lay above the CL, the data met the criteria for multiply controlled challenging behaviour (tangible- and automatically reinforced).

Figure 5.28. Matt. Percentage of intervals with other challenging behaviours during functional analysis

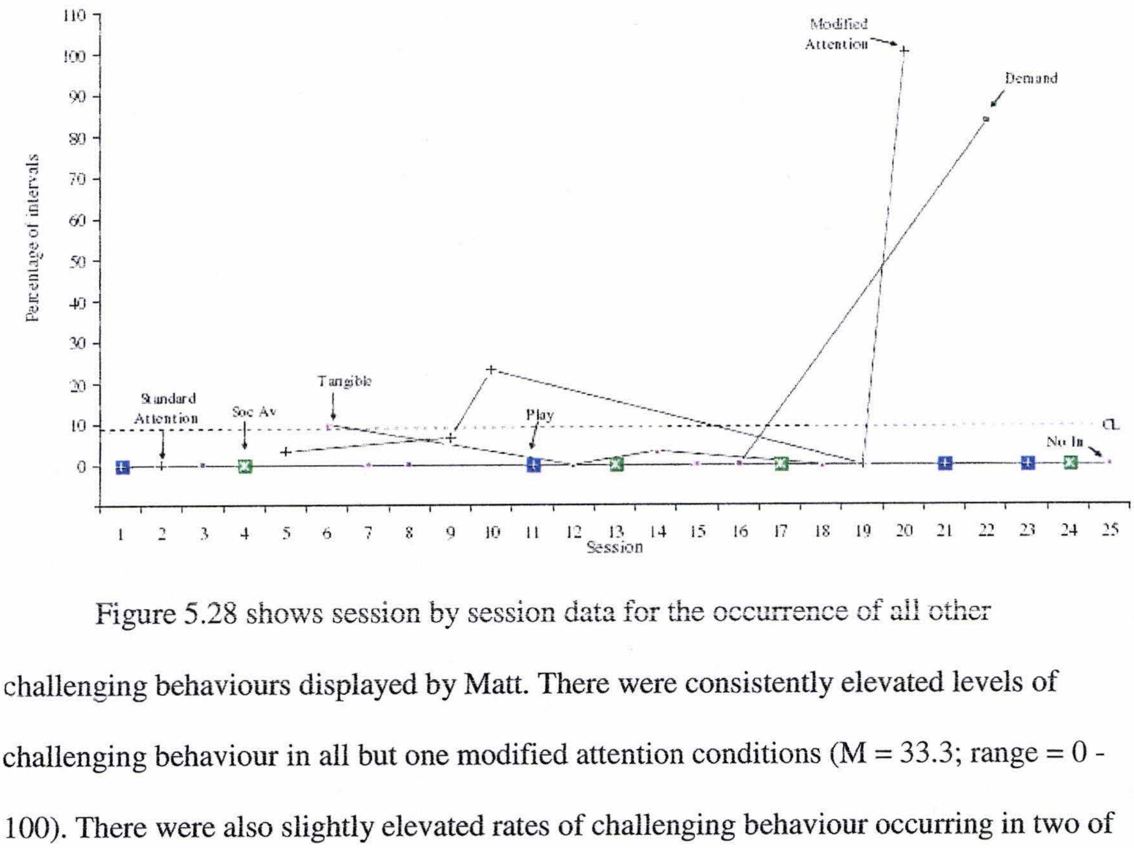


Figure 5.28 shows session by session data for the occurrence of all other challenging behaviours displayed by Matt. There were consistently elevated levels of challenging behaviour in all but one modified attention conditions (M = 33.3; range = 0 - 100). There were also slightly elevated rates of challenging behaviour occurring in two of the four tangible conditions (M = 3.3; range = 0 - 10) and a single demand condition (M = 20.8; range = 0 - 83.3). There were no occurrences of other challenging behaviours in the play, no interaction, social avoidance, or standard attention condition of the functional analysis. As with Sean, there were notable increases in the frequency, episodic severity and

[33] "In cases in which more than one condition meets criteria for differentiation, score the analysis as multiply maintained… If there are two differentiated conditions and the alone is the lower of the two, score it as both automatic and the other condition." (see Hagopian et al., 1997, p. 325).

number of topographies of Matt's challenging behaviour as the functional analysis progressed. As most of the data points were low with a small number of high data points the rules for low rate behaviours were followed[34]. As most of the high data points occurred in the modified attention condition, this was considered to be differentiated.

In sum, data for Matt suggested that object banging may have been multiply controlled by both its automatic consequences and social influences (i.e., access to tangibles). Other challenging behaviours displayed by Matt were primarily maintained by access to social contact and were specifically evoked at times in which he was asked to independently complete academic work. It may be that the presence of the demand established the reinforcing value of attention by functioning as a transitive conditioned establishing operation (CEO-T). As a parallel to the slotted screw example provided by Michael (1982), one could envisage that the onset of a demand that Matt couldn't independently complete (= the sight of the slotted screw), established social contact (= the screwdriver) as an effective type of reinforcement and evoked challenging behaviour that has led to attention in the past (= manding for the screwdriver). Indeed McGill (1999, p. 402) suggested that if demands were to function as a CEO-T for attention-maintained challenging behaviour then one would expect higher levels of such behaviour in the presence of demands than in the standard attention condition, a pattern of responding which corresponds to that reported above.

[34]"In cases in which most of the data points are low, the condition in which all or most of the higher rate behavior occurs is considered to be differentiated.... However, one of those high points must occur in the last half of the assessment." (see Hagopian et al., 1997, p. 325).

For Matt, there were a total of 40 intervals in the modified attention condition in which challenging behaviour occurred, in comparison to 25 intervals in the demand condition and 4 intervals in the tangible condition.

Angus.

Angus was a boy aged ten years old with a diagnosis of SMS. Angus had good receptive and expressive communication skills. Angus attended a school for children with severe learning disabilities and lived in the family home. The QABF was completed as part of study one by Angus' mum and this indicated that challenging behaviours appeared to be multiply controlled. Angus' challenging behaviours were also reported to be influenced by his sleep cycle and he scored highly on the physical discomfort related-subscale of the QABF. The total subscale scores for Angus are presented in Table 5.13.

Table 5.13

Angus. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
Angus.			
Attention	6	14	14
Tangible	9	14	10
Demand	3	14	15
Physical discomfort	15	15	15
Automatic	7	0	0

Target behaviours included aggression, property destruction, self-injury (banging head with his hand, chin-banging, teeth-banging, faux self-injury, finger-biting and object mouthing) and ear covering. There were no occurrences of property destruction or aggressive behaviours in the functional analysis. Target behaviours were collapsed into finger biting/object mouthing and other challenging behaviours. Inter-rater reliability was calculated for 32% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for finger biting/object mouthing was 88.9% and for other challenging behaviours was 81.8%.

The functional analysis was conducted in a large room at school. The demand condition involved the use of a series of arithmetic worksheets. The tangible condition involved withholding access to preferred comics. Following discussion with Angus' mother a single diverted attention condition was used in the initial visit, however due to the non-occurrence of challenging behaviour this was not used again. All conditions were run in accordance with the study protocol.

Results of the functional analysis suggested that challenging behaviour formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.29. Angus. Percentage of intervals with finger biting and object mouthing during functional analysis.

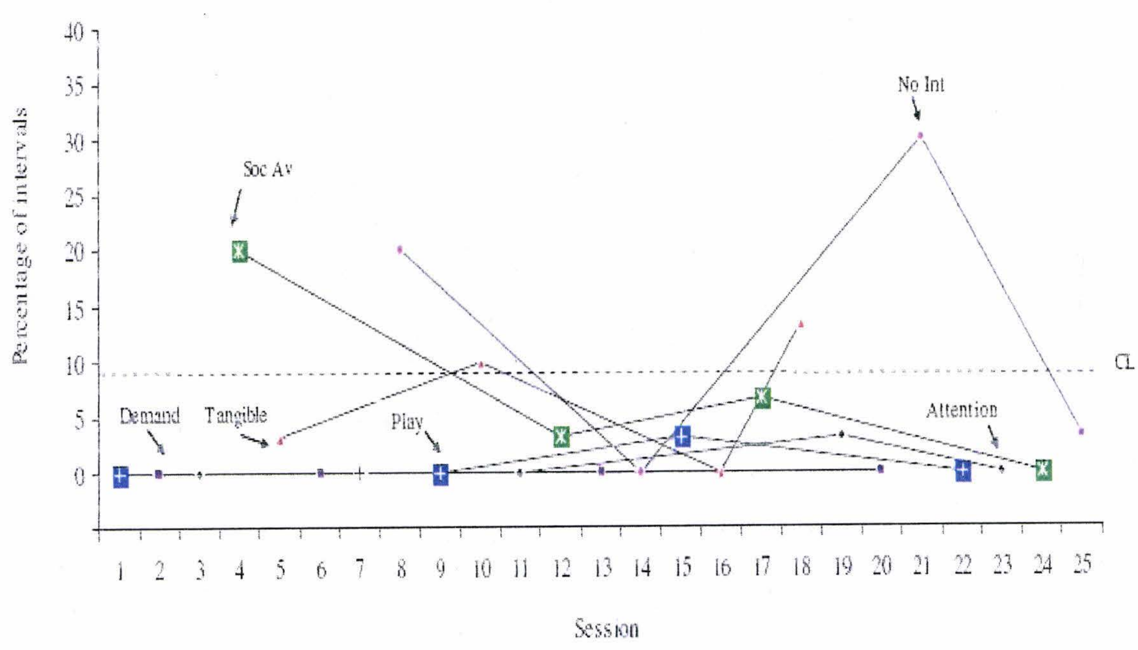


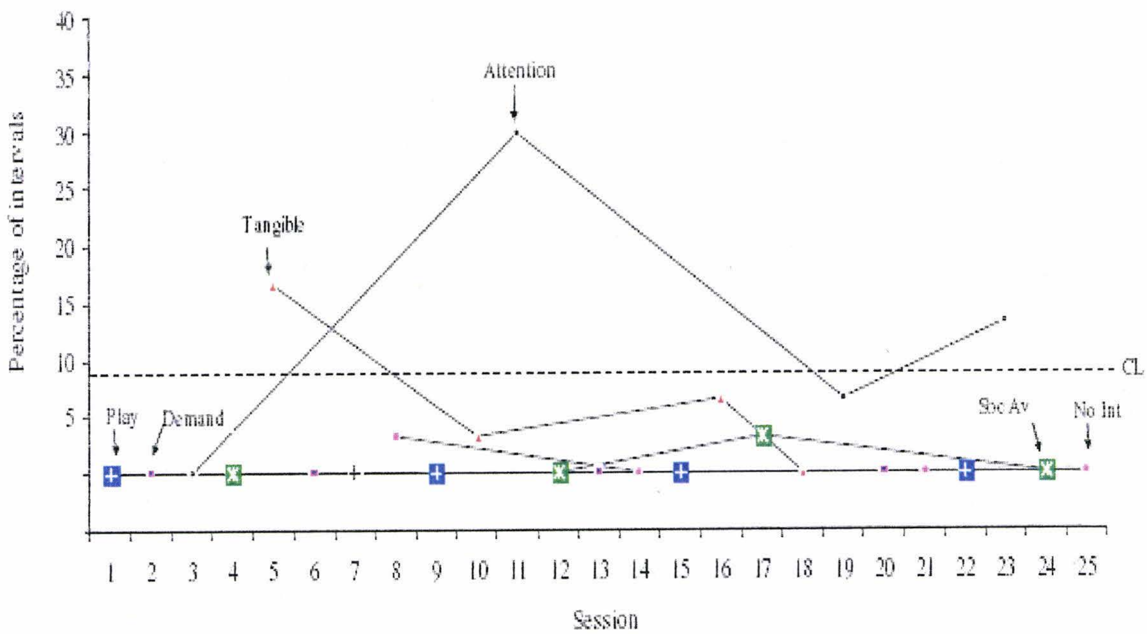
Figure 5.29 shows session by session data for the occurrence of finger biting and object mouthing. There were elevated levels of these behaviours in tangible (M = 6.7; range = 0 - 13.3), social avoidance (M = 7.5; range = 0 - 20) and no interaction conditions

(M = 13.3; range = 0 - 30). There were low rates of these behaviours in the attention and play conditions (M = .83; range = 0 - 3.3) and no occurrences in the demand condition.

As 50% of scores for the no interaction condition lay above the CL the behaviour met the criteria for automatically reinforced behaviour. No other condition met these criteria.

Figure 5.30 shows session by session data for the occurrence of all other challenging behaviours displayed by Angus. There were consistently elevated levels of challenging behaviour in all but one attention conditions (M =12.5; range = 0 - 30). There were also elevated rates of challenging behaviour occurring in three of the four tangible conditions (M = 6.5; range = 0 - 16.6). There was a single occurrence of challenging behaviour in both the no interaction and social avoidance condition (M = .83; range = 0 - 3.3). There were no occurrences of other challenging behaviours in the play or demand conditions.

Figure 5.30. Angus. Percentage of intervals with other challenging behaviours during functional analysis.



As 50% of scores for the attention condition lay above the CL the behaviour met the criteria for attention-maintained behaviour. No other condition met these criteria.

In sum, data for Angus suggested that finger biting and object mouthing may have been primarily automatically reinforced. Other topographies of challenging behaviour appeared to form an alternative response class and were primarily attention-maintained.

Patrick.

Patrick was a boy aged eleven years old with a diagnosis of SMS. Patrick had good receptive and expressive communication skills and could clearly make his needs known. Patrick attended an independent school for children with disabilities and lived in the family home. The QABF was completed as part of study one by Patrick's father and this indicated that both aggression and property destruction appeared to be at least in part attention-maintained. No clear function was identified for self-injurious behaviours using the QABF. The total subscale scores for Patrick are presented in Table 5.14.

Table 5.14

Patrick. Total subscale scores from QABF subscales.

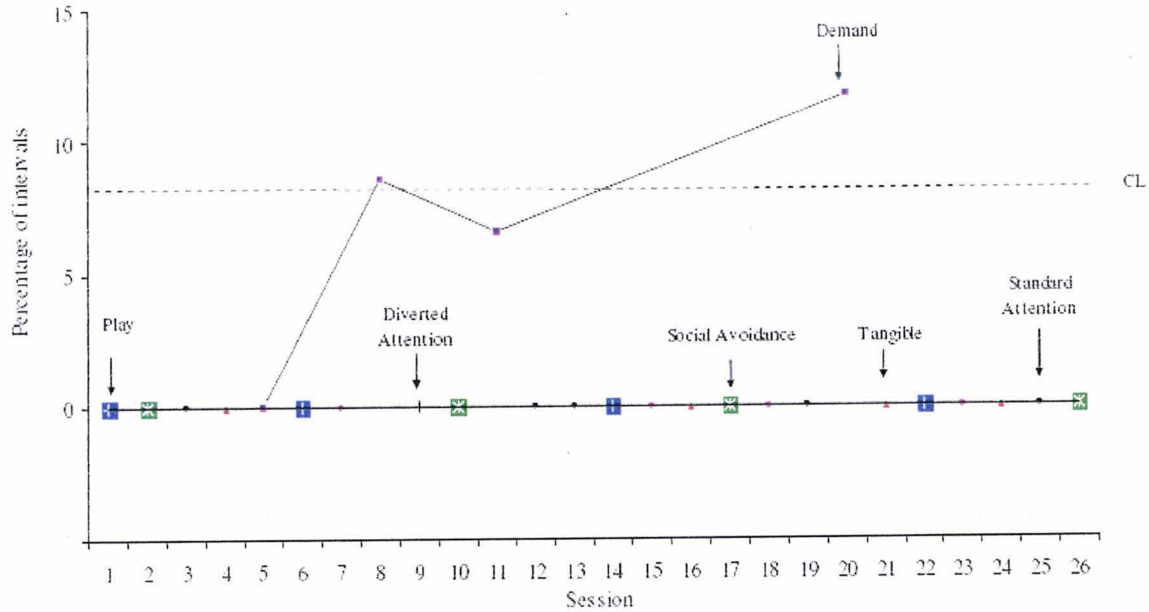
	Self-injurious behaviour	Aggression	Property Destruction
Patrick.			
Attention	6	12	12
Tangible	5	6	4
Demand	7	8	6
Physical discomfort	1	3	0
Automatic	8	2	10

Target behaviours included aggression (head butting, biting, pinching, scratching), self-injury (banging head on surfaces, tapping head with fingers or objects), property destruction (dismantling objects). The only behaviours to occur during the analysis were tapping of the head with fingers or objects (such as the nib of a pen); this often resulted in a red mark on the area where Patrick had been tapping. Target behaviours were collapsed and coded as a single response. Inter-rater reliability was calculated for 34.6% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability was 85.7%.

The functional analysis was conducted in a relatively large room at school. The demand condition involved the use of a series of arithmetic worksheets selected by Patrick's teacher. To better reflect academic tasks in Patrick's natural classroom context, session lengths varied in the demand condition, lasting anywhere between 5-10mins, and continued until the worksheet was finished. The tangible condition involved withholding access to preferred balloons. Following discussion with Patrick's father a single diverted attention condition was used in the initial visit, however due to the non-occurrence of challenging behaviour this was not used again. All other conditions were run in accordance with the study protocol.

Figure 5.31 shows the results of the functional analysis for Patrick. There were elevated rates of challenging behaviour solely in the demand condition of the functional analysis (M= 6.7; range = 0 - 11.7). There were zero occurrences of challenging behaviour in any other condition of the functional analysis.

Figure 5.31. Patrick. Percentage of intervals with challenging behaviours during functional analysis.



As 50% of scores for the demand condition lay above the CL the behaviour met the criteria for escape-maintained behaviour. No other condition met the criteria for differentiation.

In sum, head tapping appeared to be primarily escape-maintained. It is unclear as to why other target behaviours, such as aggression, were not observed during the functional analysis. These occurred at relatively high levels outside of the confines of the experimental analysis. It is possible that the behaviour was in part governed by verbal rules making Patrick insensitive to the contingencies of the functional analysis. Patrick would, for example, frequently tact that he was being a 'good boy' throughout the session.

Shelley.

Shelley was a nine year old girl with a diagnosis of Smith-Magenis syndrome. Shelley had a hearing impairment and though non-verbal was fluent in British Sign Language (BSL). Shelley attended an independent school for children with disabilities and lived in the family home. No QABF data was available for Shelley as her mother opted not to take part in study one.

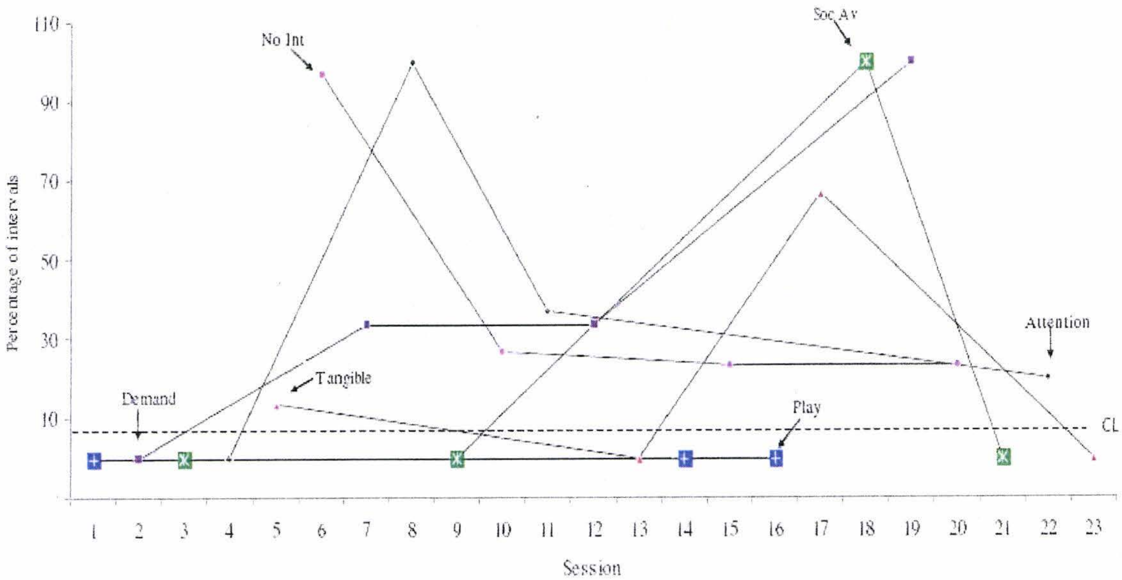
Target behaviours included aggression (lashing out with arm or elbow in proximity of the therapist), self-injury (banging head on surfaces, banging fists/legs against surfaces), property destruction (dismantling hearing aid) and non-compliance (lying on floor, sitting on arms). Shelley also inserted objects into bodily orifices; however, due to concerns regarding the potential risks of including this behaviour in the functional analysis, all such behaviours were ignored or if necessary blocked during the functional analysis. Target behaviours were collapsed and coded as either lying on the floor or other challenging behaviours. Inter-rater reliability was calculated for 34.7% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for lying on the floor was 95.5% and for other challenging behaviours was 80.9%.

The functional analysis was conducted in a large room at school. The demand condition involved the use of a series of worksheets prepared by Shelley's teacher. The tangible condition involved withholding access to a spinning top toy. All conditions were run in accordance with the study protocol.

Results of the functional analysis suggested that challenging behaviour formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.32 shows session by session data for the occurrence of lying on the floor. There were elevated levels of these behaviours in the no interaction condition (M = 42.5; range = 23.3 – 96.7), demand condition (M = 41.7; range = 0 - 100), attention condition (M = 39.2; range = 0 - 100) and two sessions of the tangible condition (M = 20, range = 0 - 66.7). There were zero rates of dropping in all but one social avoidance condition (M = 25, range = 0 - 100) and in all play conditions.

Figure 5.32. Shelley. Percentage of intervals with lying on floor during functional analysis.

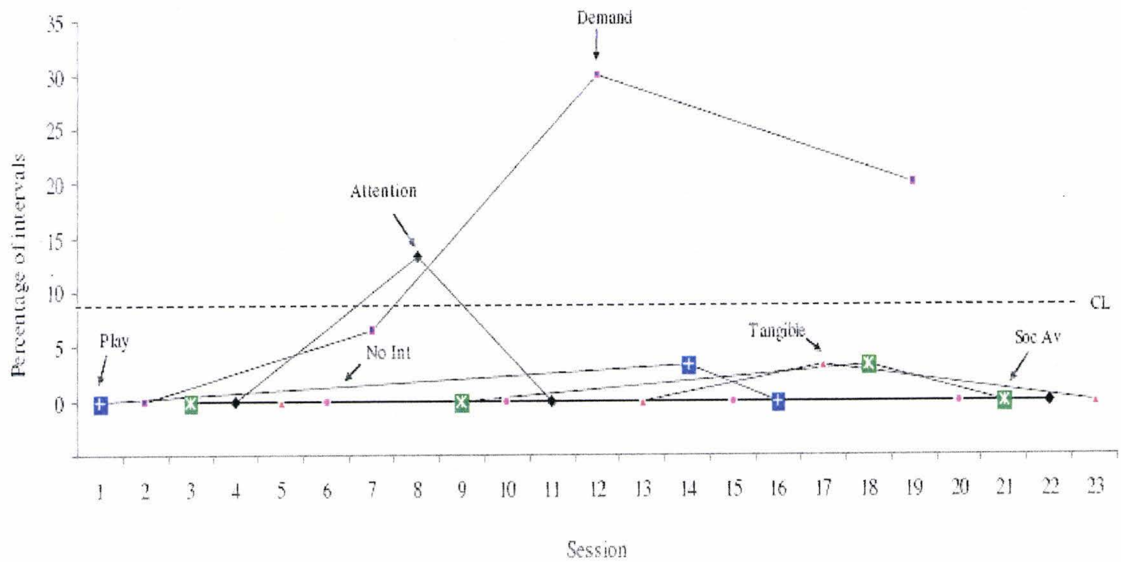


As multiple conditions met criteria for differentiation the Hagopian et al rules for multiple maintaining variables were adopted[35]. As the highest rate of responding occurred in the no interaction condition lying on the floor met the criteria for automatically reinforced behaviour. It should be noted, however, that the no interaction condition shares the same antecedents as the attention condition and that high levels of lying on the floor may have been an artefact of this experimental arrangement. As such, whilst lying on the floor met criteria for automatically reinforced behaviour it is difficult to completely rule out a multiply socially controlled hypothesis.

Figure 5.33 shows session by session data for the occurrence of other challenging behaviours. There were consistently elevated levels of these behaviours in the demand condition (M = 14.2; range = 0 – 30). There were elevated levels of other challenging behaviours in a single attention condition (M = 3.3; range = 0 - 13.3). There were zero levels of challenging behaviour in all other experimental conditions.

[35] "In cases in which more than one condition meets criteria for differentiation, score the analysis as multiply maintained (unless the highest is alone; then score it *only* as automatic)." Hagopian et al (1997, p. 325).

Figure 5.33. Shelley. Percentage of intervals with other challenging behaviours during functional analysis



As 50% of data points in the demand condition fell above the CL this condition met the criteria for escape-maintained behaviour.

In sum, data for Shelley suggested that challenging behaviours formed two separate response classes. Lying on the floor met the criteria for automatically reinforced behaviour (although a multiply socially controlled account could not be completely ruled out). Other challenging behaviours appeared to be escape-maintained. As with both Matt and Sean there was a notable increase in the episodic severity of challenging behaviours as the functional analysis progressed.

Katie.

Katie was an eleven year old girl with a diagnosis of Smith-Magenis syndrome. Katie had good expressive and receptive communication skills and could clearly make her needs known verbally. Katie attended an independent school for children with disabilities and lived in the family home. The QABF was conducted with Katie's classroom teacher as part of study one. The results of the QABF suggested that both aggression and property destruction were primarily escape maintained. The total subscale scores for Katie are presented in Table 5.15.

Table 5.15

Katie. Total Subscale Scores from QABF Subscales.

	Self-injurious behaviour	Aggression	Property Destruction
Katie.			
Attention	N/A	7	9
Tangible	N/A	4	4
Demand	N/A	9	11
Physical discomfort	N/A	2	2
Automatic	N/A	4	[illegible]

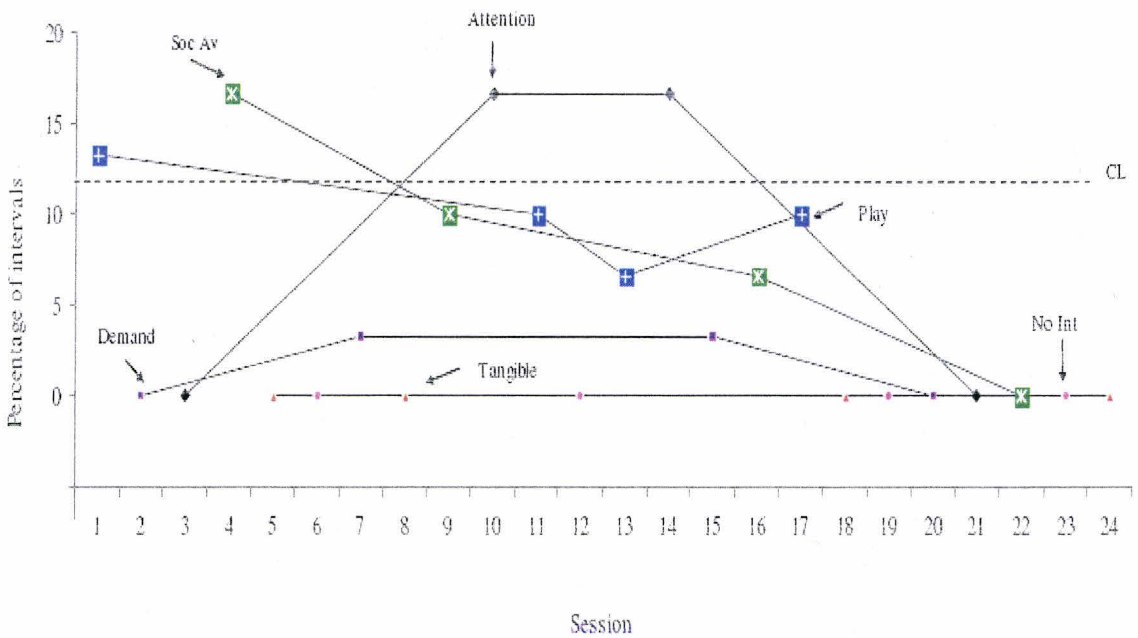
Target behaviours included self-injury (self-slapping, repetitive banging of face with objects), property destruction (breaking pens), aggression (nipping, kicking, hair pulling). Katie also displayed a number of stereotypical behaviours, such as the spasmodic upper body squeeze reported as being characteristic of Smith-Magenis syndrome (Finucane, Konar, Haasgivler, Kurtz, & Scott, 1994). As such stereotypical behaviours (spasmodic upper body squeeze, lifting both hands to the side of face and extending fingers) were also included as target behaviours in the functional analysis. There were no occurrences of aggressive behaviours or property destruction in the functional analysis and it was reported by Katie's teacher that these behaviours were typically only directed at other children. Target behaviours were collapsed and coded as either stereotypical behaviour or self-injurious behaviours. Inter-rater reliability was calculated for 33.3% of experimental sessions. Using the more stringent measure of reliability (R-Occ) the reliability for stereotypical behaviours was 95% and for self-injurious behaviours was 100%.

The functional analysis was conducted in a large room at school. The demand condition involved the use of a series of worksheets prepared by Katie's teacher. The tangible condition involved withholding access to preferred comics. All conditions were run in accordance with the study protocol.

Results of the functional analysis suggested that challenging behaviour formed two separate response classes. Results of the functional analysis for each response class are therefore presented separately.

Figure 5.34 shows the results of the functional analysis for stereotypical behaviours. There were notably elevated levels of stereotypical behaviour occurring in the play (M= 9.9; range = 6.6 - 13.3), social avoidance (M = 8.3; range = 0 - 16.6) and attention (M = 8.3; range = 0 - 16.6) conditions of the functional analysis. There were low levels of stereotypical behaviour in two sessions of the demand condition (M = 1.7; range = 0 - 3.3) and zero rates of the behaviour in tangible and no interaction conditions of the functional analysis.

Figure 5.34. Katie. Percentage of intervals with stereotypical behaviours during functional analysis.



The behaviour met criteria for attention-maintained behaviour with 50% of data points in the attention condition falling above the CL. Some caution needs to be taken with this interpretation, however. The high levels of stereotypical behaviour occurring in the play condition led to the CL being set unusually high. Anecdotally the response appeared to occur during conversation and at times when the therapist commented on Katie's play. The response occurred at higher levels in the social avoidance, play and attention conditions, which all involved some commentary on Katie's play. The behaviour occurred at near zero levels in those conditions in which this source of stimulation was absent. Other studies have shown that the onset of social interaction in the play condition can evoke stereotypical behaviours by acting as an EO for automatically reinforced behaviours (Van Camp et al., 2000). The clear downward trend in the social avoidance condition would suggest that the response did not occur to escape from social attention lending support to this hypothesis.

Figure 5.35. Katie. Percentage of intervals with self-injurious behaviours during functional analysis.

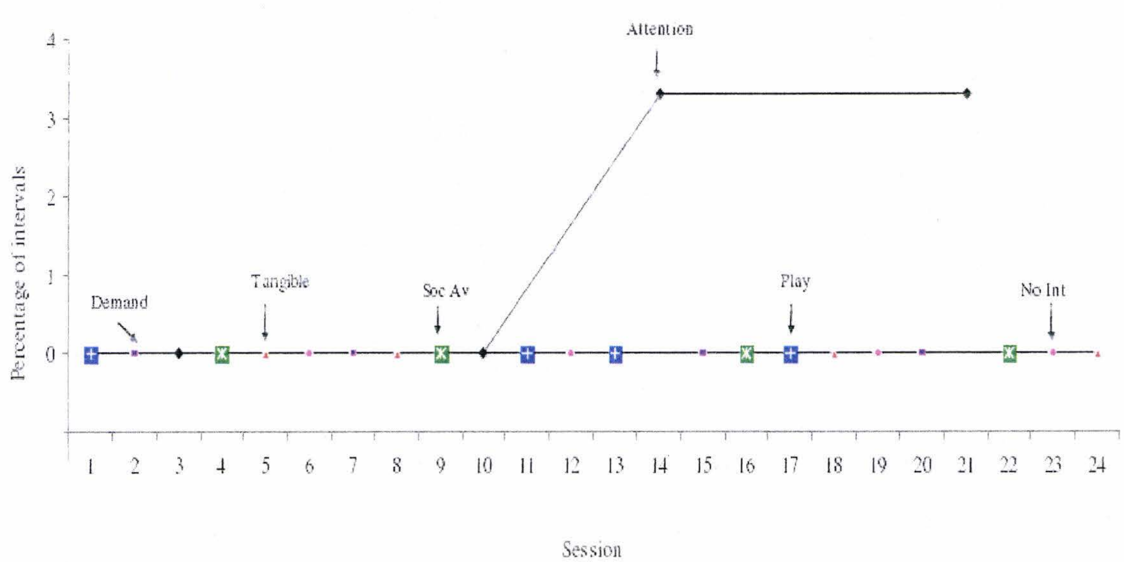


Figure 5.35 shows the results of the functional analysis for self-injurious behaviours. Self-injurious behaviours occurred in two of the four attention conditions (M = 1.65; range = 0 - 3.3). There were zero rates of the behaviour in all other conditions.

Due to the low rate of self-injurious behaviours, a CL could not be established and the Hagopian et al rules for the analysis of low-rate behaviours were utilised[36]. Accordingly self-injurious behaviour met the criteria for attention-maintained behaviour.

In sum, Katie was found to display attention-maintained self-injurious behaviour. Findings for stereotypical behaviours were relatively unclear. Whilst stereotypical behaviours met the criteria for attention-maintained behaviour, this may have been an artefact of relying on the Hagopian et al criteria for differentiation (which assumes the play condition acts as a control). There were consistently high levels of the same behaviours occurring in the play condition and social avoidance condition and further analysis is required to test the hypothesis that social interaction may have acted as an EO for the automatic consequences that may maintain this response.

Participants with SMS. Within-Group Analysis

Figures 5.36-5.42 provide summary data for each participant in the SMS group. The numerical data in each figure indicates the overall mean percentage of intervals of challenging behaviour and the overall standard deviation. The data for each experimental condition is also presented graphically to present the number of standard deviations each condition mean is from the overall mean (i.e., the Z score for each experimental condition).

In what follows, particular emphasis is given to the analysis of those topographies of challenging behaviour that appear to be socially maintained. All topographies that

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[36] "In cases in which most of the data points are low, the condition in which all or most of the higher rate behavior occurs is considered to be differentiated.... However, one of those high points must occur in the last half of the assessment." (see Hagopian et al., 1997, p. 325).

For Katie, there were a total of 2 intervals in the attention condition in which self-injurious behaviours occurred. No challenging behaviours occurred in any other condition.

appeared to be automatically reinforced were excluded from the current analysis. Stereotypical behaviours displayed by Katie, which met criteria for attention-maintained behaviour, were also excluded from this analysis due to the failure to rule out the competing hypothesis that the behaviours were automatically reinforced. A number of apparent patterns in the data are discussed.

Figures 5.36-5.39 present summary data for four participants with SMS for whom one response class of challenging behaviour was, at least in part, positively reinforced by the contingent provision of social attention. Figures 5.40-5.42 present summary data for the three participants with SMS who displayed at least one response class of challenging behaviour that appeared to be either negatively reinforced by the removal of academic demands or positively reinforced by access to tangibles.

Figure 5.36. Sean. No. of standard deviations each condition mean was from the overall mean for other challenging behaviours.

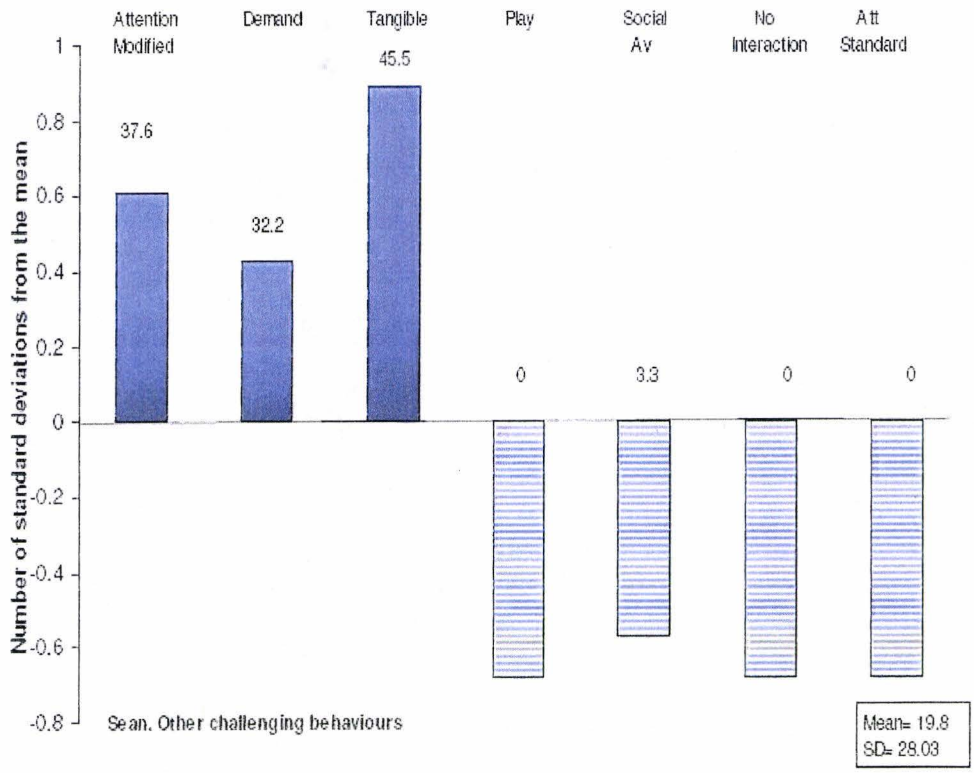


Figure 5.37. Matt. No. of standard deviations each condition mean was from the overall mean for other challenging behaviours.

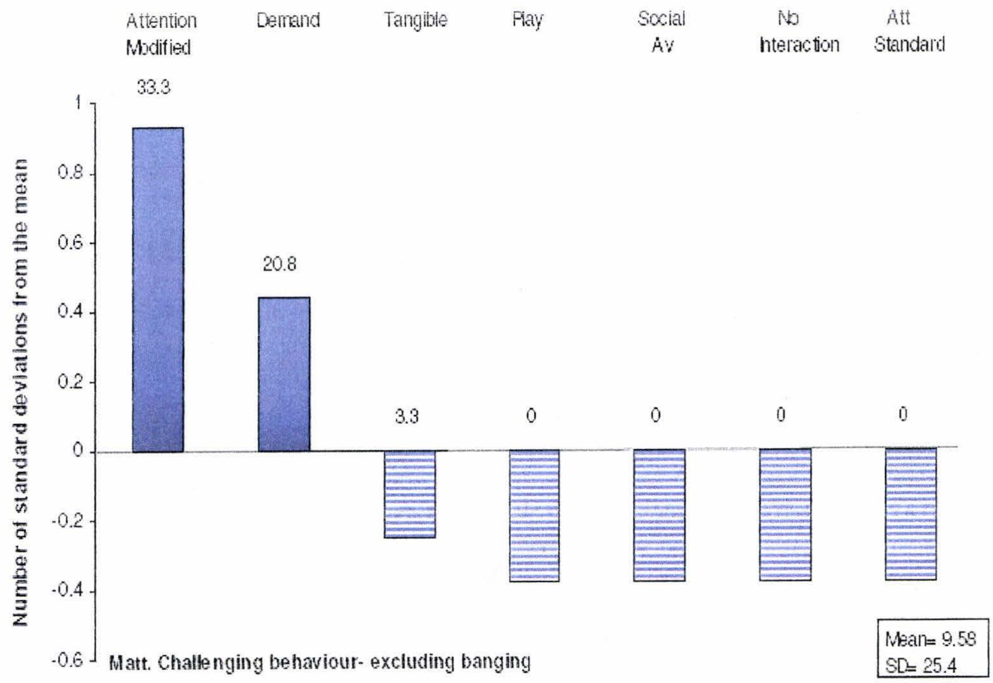


Figure 5.38. Angus. No. of standard deviations each condition mean was from the overall mean for other challenging behaviours.

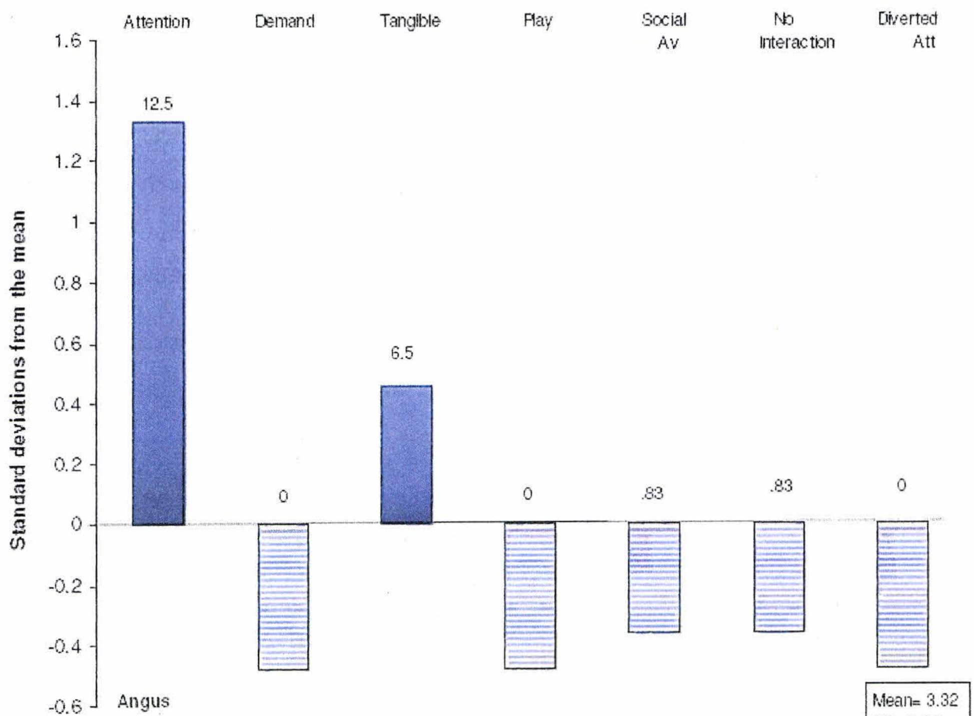
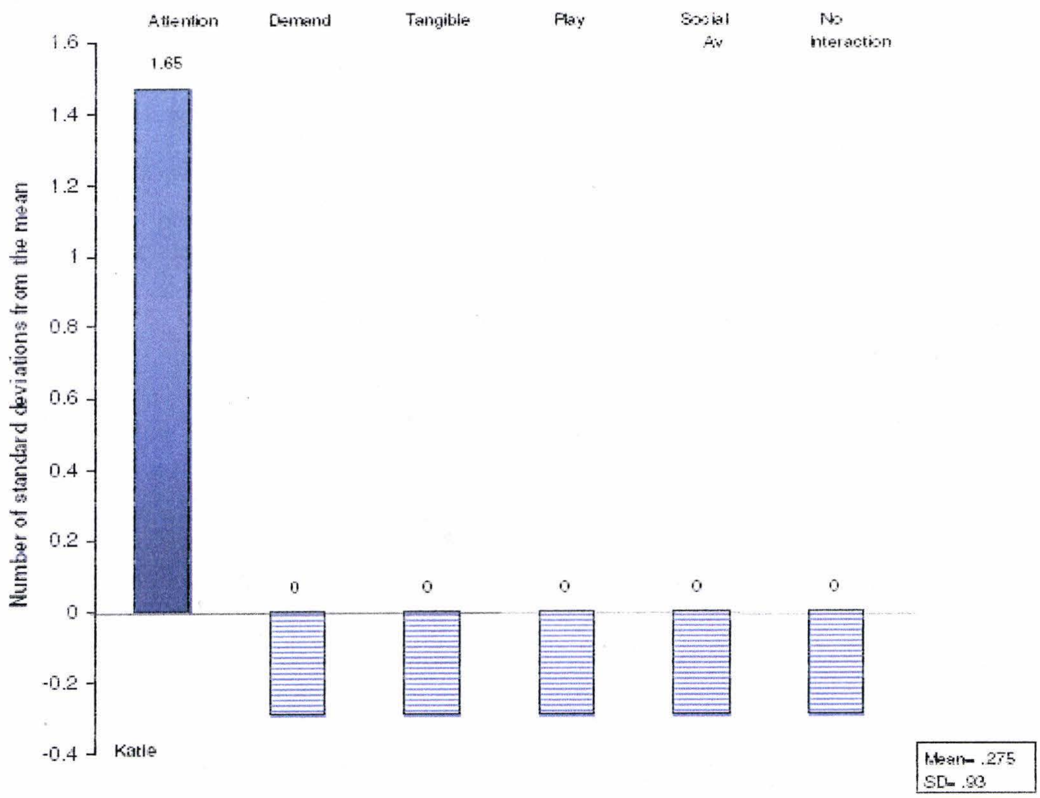


Figure 5.39. Katie. No. of standard deviations each condition mean was from the overall mean for self-injurious behaviours.



For Sean, as shown in Figure 5.36, the mean occurrence of challenging behaviour in the Tangible condition was 45.5, which was .89 standard deviations above the overall mean of 19.8. The mean occurrence of challenging behaviour in the Modified Attention condition was 37.6, which was .61 standard deviations above the overall mean. The mean occurrence of challenging behaviour in the Demand condition was 32.2, which was .43 standard deviations above the overall mean. This supports the interpretation that Sean displayed challenging behaviour that served multiple functions, one of which was attention-maintained.

For Matt, as shown in Figure 5.37, the mean occurrence of other challenging behaviours in the Modified Attention condition was 33.3, which was .93 standard deviations above the overall mean of 9.58. The mean occurrence of other challenging behaviours in the Demand condition was 20.8, which was .44 standard deviations above the overall mean. It should be remembered however that there was only a single Demand condition in which any challenging behaviours occurred. Overall this supports the interpretation that other challenging behaviours displayed by Matt were primarily attention-maintained.

For Angus, as shown in Figure 5.38, the mean occurrence of challenging behaviour in the Attention condition was 12.5, which was 1.33 standard deviations above the overall mean of 3.32. The mean occurrence of challenging behaviour in the tangible condition was 6.5, which was .46 standard deviations above the overall mean. This supports the interpretation that Angus displayed challenging behaviour that primarily served an attention function.

For Katie, as shown in Figure 5.39, the mean occurrence of challenging behaviour in the Attention condition was 1.65, which was 1.47 standard deviations above the overall mean of .275, suggesting that self-injurious behaviours were attention maintained.

Figure 5.40. Matt. No. of standard deviations each condition mean was from the overall mean for object banging.

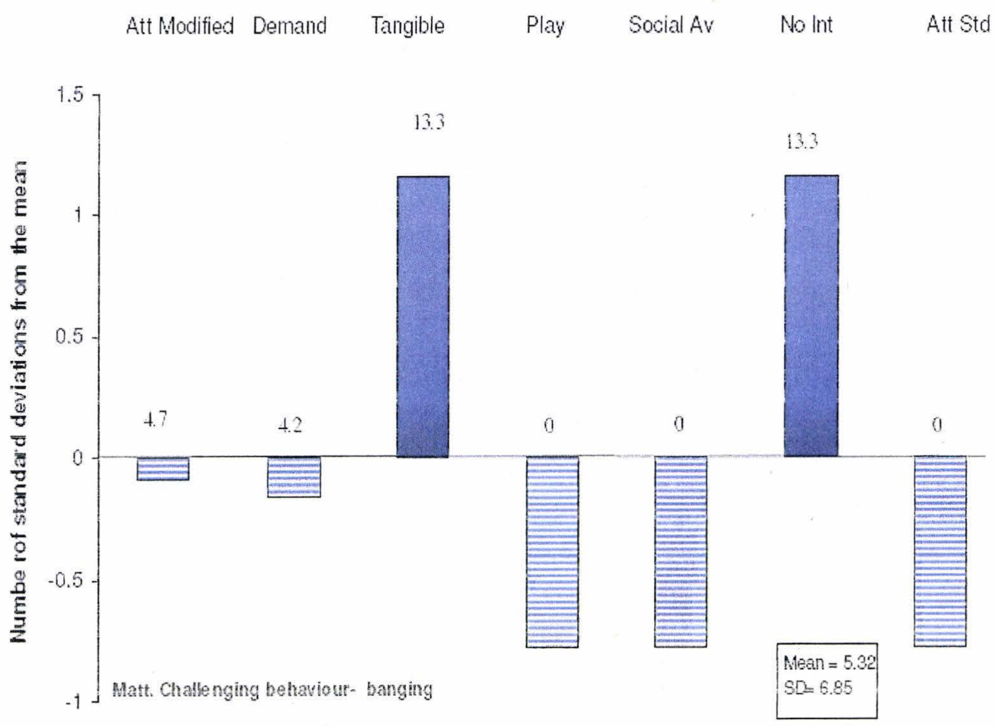


Figure 5.41. Patrick. No. of standard deviations each condition mean was from the overall mean for head-tapping.

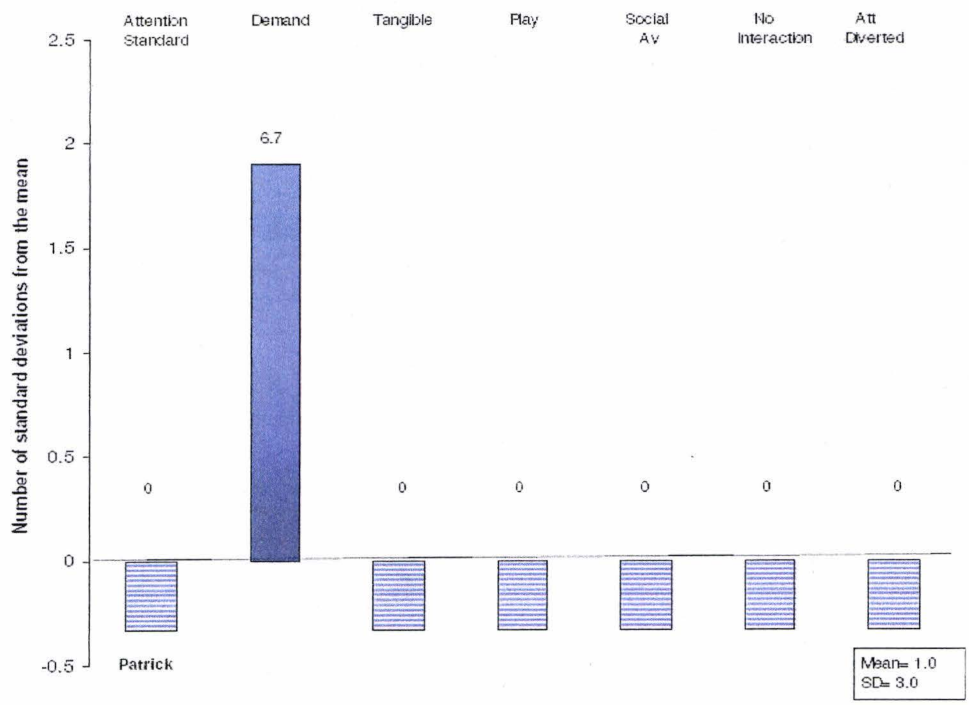
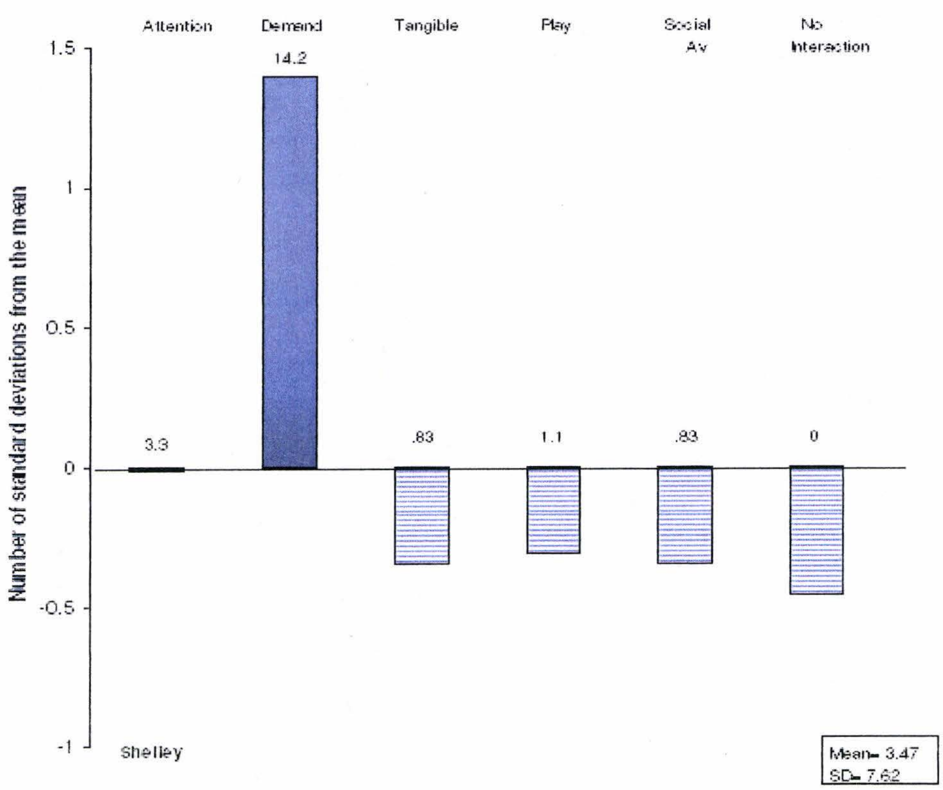


Figure 5.42. Shelley. No. of standard deviations each condition mean was from the overall mean for other challenging behaviours.



For Matt, as shown in Figure 5.40, the mean occurrence of object banging in the Tangible and the No Interaction conditions was 13.3, which was 1.16 standard deviations above the overall mean of 5.32. This supports the interpretation that object banging served multiple functions (both tangible- and automatically-maintained). For Patrick, as shown in Figure 5.41, the mean occurrence of challenging behaviour in the demand condition was 6.7, which was 1.90 standard deviations above the overall mean of 1.0. This supports the interpretation that Patrick displayed challenging behaviour that was maintained by escape from demands. For Shelley, as shown in Figure 5.42, the mean occurrence of challenging behaviour in the demand condition was 14.2, which was 1.4 standard deviations above the overall mean of 3.47. This supports the interpretation that Shelley displayed challenging behaviour that was maintained by escape from demands.

In sum, analysis of within-group variation for participants with SMS suggested two patterns of results. First a relatively high proportion of participants (4/6) displayed one response class of challenging behaviour that appeared to be, at least in part, attention-maintained. Second, 3/6 of participants with SMS displayed at least one response class of challenging behaviour that was not influenced by social attention and was maintained by either escape- or access to tangibles.

Correspondence Between Indirect and Experimental Functional Assessment Methods.

Table 5.16 shows the correspondence between the results of indirect and experimental functional assessment methods for each participant who took part in study 2 (with the exception of Shelley for whom no indirect data were collected). The correspondence between the different methodologies was relatively limited. There was no agreement between indirect and experimental methods for 3 participants (Calum, Patrick and Katie). There was partial agreement for 7 participants (Abe, Greg, Luke, John, Sean, Matt and Angus). Experimental analyses were successful in identifying a behavioural function for 3 participants in which the QABF had been unable to ascribe a function.

Table 5.16. *Correspondence Between Results of Indirect Functional Assessment and Experimental Functional Analysis Methodologies.*

Participant	QABF*	Experimental functional analysis**
FXS		
Abe	Tangible, Escape, Automatic	Tangible, Automatic
Greg	Escape, Automatic	Escape (social avoidance), Tangible
Jacob	Function not identified	Escape (demand)
Luke	Escape, Tangible	Escape (demand)
Theo	Function not identified	Escape (demand)
John	Escape, Tangible, Automatic	Escape (demand)
Calum	Escape, Automatic	Tangible
Richard	Function not identified	Tangible
SMS		
Sean	Attention, Tangible, Escape	Attention, Tangible, Escape (demand), Automatic
Matt	Attention, Escape	Attention, Tangible, Automatic
Angus	Attention, Tangible, Escape	Attention, Automatic
Patrick	Attention, Automatic	Escape (demand)
Katie	Escape	Attention

* For a function to be ascribed using the QABF there had to be a score of 10 or more on the relevant subscale (necessitating that 4 of the 5 items for the subscale were endorsed). Data from the Physical discomfort related subscale were excluded as this function was not assessed in the experimental functional analyses.

**As there is no specific social avoidance subscale on the QABF, social avoidance and demand functions identified via experimental functional analyses were collapsed into the category 'Escape'.

Social Acceptability.

A total of ten stakeholders returned completed assessment of social acceptability forms. Table 5.17 shows summary statistics for each item of the assessment.

Table 5.17

Social Acceptability of Analogue Assessments. Results of Acceptability Questionnaire.

Item: Paraphrased Question	Mean	Range
All items scored 1 (strongly disagree) – 5 (strongly agree)		
1. I find this assessment acceptable.	3.8	(2-5)
2. I would use this assessment again	4.3	(4-5)
3. It would be acceptable to use this assessment without a child's consent	3.6	(2-5)
4. I like the procedures used in the assessment	4.1	(3-5)
5. The assessment is likely to be effective in identifying the function of the child's behaviour	3.9	(3-5)
6. I believe the child experienced discomfort during the assessment	2.5	(1-4)
7. I believe the assessment will result in permanent improvement in the child's behaviour	2.9	(2-4)
8. I believe it is acceptable to use this assessment with people who lack capacity to consent	3.6	(2-4)
9. Overall I had a positive reaction to this assessment.	4.3	(3-5)

The key items from the perspective of the current study were items 1, 6 and 9. In response to item 1, stakeholders generally found the assessment acceptable (M = 3.8; range = 2-5). In response to item 6, the majority of stakeholders did not believe the child had experienced discomfort as a result of the assessment (M = 2.5; range = 1-4). Finally, in response to item 9, overall stakeholders reported having a positive reaction to the assessment procedures adopted in the current study (M = 4.3; range = 3-5).

Summary of Results

Individual variation

- There was considerable individual variation in the occurrence of challenging behaviours for each participant across each analogue condition.
- All participants displayed at least one response class of challenging behaviour that was socially-influenced.
- In some cases the standard analogue protocol (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982) required individualisation to capture relevant antecedent and consequence relations.

Within group

FXS

- No child with FXS displayed any response class of challenging behaviour that appeared to be attention-maintained.
- Four children with FXS displayed one response class of challenging behaviour that appeared to be tangible-maintained.
- Five children with FXS displayed one response-class of challenging behaviour that appeared to be negatively reinforced by either the removal of demands or social attention.

SMS

- Four of the six SMS participants displayed one response class of challenging behaviour that was, at least in part, attention-maintained.
- Three of the six children with SMS displayed one response class of challenging behaviour that was not influenced by social attention and was maintained by either access to tangibles or escape from demands.

Between group

- The groups appeared to differ only in relation to the likelihood of displaying attention-maintained challenging behaviour.
- Children in both groups were shown to display escape- and tangible-maintained challenging behaviours.

Social acceptability

- The assessment methods were rated as acceptable by key stakeholders.

Discussion

All children who took part in the current study were found to display challenging behaviour that served social and non-social functions. These findings suggest that challenging behaviours in FXS and SMS rather than being genetically determined, are influenced by the same behaviour-environment relations that are known to influence challenging behaviour in people with intellectual and developmental disability generally.

The current study also revealed both within- and between-group differences in the function served by challenging behaviour in FXS and SMS. No child with FXS was found to display attention-maintained challenging behaviour; four children with FXS were found to display tangible-maintained challenging behaviour and five were found to display negatively reinforced challenging behaviour. In contrast, four of the six children with SMS were found to display challenging behaviours that were, at least in part, attention-maintained challenging behaviour and three children displayed response classes of challenging behaviour that were not influenced by social attention. When considered in the light of the findings of the study presented in the previous chapter and the existing literature, these results appear to provide further evidence for a potential gene-environment interaction (GxE) in challenging behaviour in FXS and SMS.

In the previous chapter, children with FXS were reported by parents and care-givers to be significantly less likely to display attention-maintained challenging behaviours than tangible- or escape-maintained challenging behaviours; and were reported to be significantly less likely to display attention-maintained challenging behaviours in comparison to other groups. A number of other empirical studies have examined behavioural function in FXS. These studies have suggested a relatively high proportion of challenging behaviour in FXS may be escape-maintained (e.g., Hall et al., 2006; Symons et al., 2003; Woodcock et al., 2009). The results of the Symons et al study also suggested elevated levels of tangible-maintained challenging behaviour and unusually low levels of attention-maintained challenging behaviour.

The results of the current study are consistent with these findings. The current study has extended this line of work by using analogue functional analysis methods, providing a 'believable demonstration' of the relations described above rather than relying on indirect or uncontrolled methods of functional assessment. In addition, the studies cited above have not allowed for a clear examination of the role played by social attention in challenging behaviour in FXS. In their study, for example, Hall et al were unable to determine whether problem behaviours in children with FXS occurred to escape from demands or social attention. Thus, it remains relatively unclear whether social attention acts as an aversive stimulus for children with FXS, or whether it simply does not function as an effective type of reinforcement. In the current study, only one child displayed differentially high levels of challenging behaviour in the social avoidance condition of the functional analysis. This suggests that generally speaking social contact did not function as an aversive stimulus for the other children in the FXS group. The low levels of challenging behaviour in the attention condition of the functional analysis for all children with FXS would seem to support the hypothesis that the value of attention as a type of reinforcement may be abolished in this particular group.

In the previous chapter, children with SMS were reported by parents and care-givers to score relatively highly across all subscales of the QABF. In comparison to other groups, children with SMS were found to be more likely to display attention-maintained and pain-related challenging behaviours. Clinical descriptions of people with SMS have repeatedly reported the apparent high 'need' such children have for adult attention (e.g., A. Smith et al., 1998). A handful of studies have examined behavioural function in SMS (Bass & Speak, 2005; Sloneem, 2005; Taylor & Oliver, 2008). These studies have suggested that challenging behaviour in SMS may be especially likely to be attention-maintained. For example, Taylor and Oliver (2008), using descriptive lag sequential analysis methods, reported that the pattern of challenging behaviour observed in four out of the five individuals with SMS in their sample was consistent with an attention-maintained hypothesis.

The findings of the current study, to some extent, appear to reflect those described above for children with SMS. For example, four children with SMS were found to display challenging behaviour that was, at least in part, maintained by social attention. This was in marked contrast to the findings for the FXS group and may provide some support for the assertion that children with SMS may be more likely to display attention-maintained challenging behaviours (Taylor & Oliver, 2008). In the current study, children with SMS were also found to display challenging behaviours that served other social and non-social functions. Whilst this is something that was also reported in the indirect findings presented in the previous chapter; other empirical work, such as that by Taylor & Oliver (2008) have not reported such relations. Judging from the present findings at least, children with SMS may be more likely to display challenging behaviour that serves multiple functions, one of which may be attention-maintained. However, it remains unclear as to whether children with SMS are more likely to display attention-maintained behaviour than any other behavioural function.

The current findings therefore seem to suggest that GxE may hold some implications for the understanding of the development and maintenance of challenging behaviour. One suggested means by which genetic events influence behaviour is by enduringly establishing (or abolishing) the reinforcing (or punishing) value of certain behavioural consequences (Kennedy et al., 2001). In this sense, it may be that certain genetic events operate as a type of motivating operation. The within- and between-group differences in behavioural function reported above for children with FXS and SMS appear to have provided some support for this hypothesis using experimental analogue methods.

It is worth reflecting on the use of analogue methods adopted in the current study as within a UK context such methods are rarely used either clinically or in research. One possible reason for the low uptake of analogue methodology in the UK may be due to ethical concerns some hold regarding the use of such methods (Hastings & Noone, 2005). The social acceptability of such assessment methods may therefore be a possible concern for clinicians and researchers alike. Key stakeholders in the current study all reported having a positive reaction to the use of analogue assessment methods and on the whole regarded them as a socially acceptable form of assessment. Only three stakeholders indicated that they felt their child experienced any discomfort as a result of the assessment. These data suggest that analogue methods may be more socially acceptable than is often assumed. To the author's knowledge these are the first data to be reported on the social acceptability of analogue methods of functional analysis modes of assessment.

There are a number of limitations to the current study that merit some consideration. There were a number of threats to the internal validity of each analogue assessment. In some cases, these could be controlled and incorporated into the assessment. For example, variations were made to the way in which demands were presented, the location in which the assessment was conducted, and the specific antecedents used in the attention condition. These manipulations all appeared to influence the occurrence of challenging behaviour for these individuals. For some individuals, however, there appeared to be uncontrolled sources of variability that may have influenced the occurrence of challenging behaviour. Three children with SMS (Sean, Matt and Shelley) showed a notable increase in the frequency and severity of challenging behaviour on particular days. Care-givers anecdotally attributed this to sleep disruption; however, a controlled analysis of such factors was beyond the scope of the current study. In addition, the no interaction condition may not have provided the most optimal of tests for automatically reinforced challenging behaviour. In this condition, the EO for attention (deprivation of attention) is present, as are salient discriminative stimuli for attention (i.e., the presence of the 'therapist'), recent work on the evocative effect of the EO (e.g., O'Reilly, Sigafoos et al., 2006) suggests that high levels of challenging behaviour occurring in this condition could potentially be indicative of an attention rather than an automatic function. That said, this interpretation could equally apply to the standard alone condition of a functional analysis (which also contains the EO for attention).

A number of individuals did not display the same topographies of challenging behaviour within the assessment that they were reported to in natural settings. A number of studies have noted differences in the results of analogue assessments depending on who is running the condition (Ringdahl & Sellers, 2000). Perhaps this underpinned the poor correspondence between the results of indirect and experimental functional assessment methodologies in the current study[37]. The internal and external validity of the assessments

[37] Although this may also have been a result of the difficulty associated with asking caregivers to correctly describe functional relations that underpin challenging behaviour. For example, parents may describe that their child's challenging behaviour occurs in the context of demands, however it can be very difficult to determine whether this occurs to escape from the demand or because other things (such as attention or access

may therefore have been improved by training parents/care-givers to run the assessment conditions and/or by using more naturalistic functional assessment methods, such as SDAs. However, there were time and resource constraints which precluded the use of the former and concerns over internal validity which precluded the use of the latter.

An analogue functional analysis should continue until there is a clear differentiation between experimental conditions (Hanley et al., 2003). However, in order to manage the practical demands of the current study, a limit of up to four visits was imposed in which to conduct the assessment. In some cases therefore, the assessment was terminated earlier than would otherwise have been the case. This either resulted in a relatively small number of replications for certain conditions or the termination of the analysis when there was still considerable variability in the data. That said, a decision was able to be reached regarding the function served by challenging behaviour for each individual.

For a number of participants, it proved to be very difficult to run the no interaction condition with any procedural integrity. For example, for some children it was difficult to contain them within a room or to prevent interaction with other family members. As such not all participants were exposed to sufficient replications of this condition.

The within- and between-group findings reported in the current study are somewhat hampered by the relatively low N and the reliance on convenience sampling. Thus, it is possible that subject variables, other than genetic status, contributed to the within- and between-group differences reported above. For this reason, study one involved a comparison involving a larger N than was possible in the current study, due to the resource intensive nature of analogue work. Future research is warranted therefore that employs larger N than was possible in the current study to examine this same research question using similar analogue methods.

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to tangibles) happen to be withheld at the same time as the demand. Functional analysis allows such competing hypotheses to be directly tested.

The methods used to assess social acceptability in the current study fell somewhat short of 'state of the art' (Schwartz and Baer, 1991). For example, the generalisability of the assessment was limited to parents of children with specific syndromes, little is known regarding the views of society at large regarding the acceptability of functional analysis methods. In addition, the accuracy of the social acceptability ratings is relatively unknown. This could have been improved by comparing ratings of the acceptability of indirect functional assessment methods against experimental counterparts and should be addressed in future research.

Despite these limitations, the current study provided some further evidence of between- and within-group differences in the function served by challenging behaviour in children with FXS and SMS. These findings may have a number of implications for future research.

First, it would be of interest to begin to examine the underpinnings of these apparent GxE. The notion of the *endophenotype* may be important in this regard. For example, FXS is associated with changes in the functioning of the L-HPA axis which is thought to mediate the stress response. It would be important for future research to begin to examine the relationship between such factors and the function served by challenging behaviour in this particular syndrome group. Similarly, it was not possible to examine the influence of health related factors on challenging behaviour in the current study, despite study one suggesting that such variables may be especially pertinent for children with SMS. Given the high levels of sleep disruption associated with the syndrome it would be of interest to begin to examine the influence of such variables more comprehensively using analogue methods.

Second, one of the major implications of the current findings lies in relation to current understanding of the development of challenging behaviour and the early intervention and prevention of such behaviours. It would be interesting to examine whether young children with SMS and FXS show differences in the way in which challenging behaviours develop and to evaluate the effects of secondary prevention efforts targeted at children from such syndrome groups.

Third, the generalizability of the findings from the current study needs to be further established. This could in part be achieved by replicating the current study with a larger N in order to examine such differences statistically. In addition, it needs to be determined whether similar GxE influence challenging behaviour displayed by individuals with other syndromes.

In sum, the findings of the current study suggest that challenging behaviour may be influenced by GxE. Specifically, no child with FXS displayed attention-maintained challenging behaviour. Children with SMS all displayed challenging behaviour that served multiple functions, one of which may have been attention. The use of analogue methods to examine this question extends the existing literature. In short, the current study has provided some experimental evidence to suggest that genetic events may serve to influence the reinforcing value of those consequences that maintain challenging behaviour.

Chapter VI

General Discussion.

"Context imbues behavior with meaning- the meaning of behavior emerges from its context. Behavior is not the mere interplay of materially defined stimuli and responses, but rather is represented by strong reciprocal interactions among stimulus and response functions in context- where those functions emerge from their context and nowhere else." (E. K. Morris, 1988, p. 309)

Chapter Overview

The findings of the current thesis suggest that there may be between-syndrome differences in the function served by challenging behaviours displayed by children with different genetic syndromes. Children with FXS and SMS differed in the likelihood of displaying attention-maintained and pain-related challenging behaviours. Such evidence supports the hypothesis that genetic events may enduringly alter the reinforcing (or punishing) value of certain behavioural consequences.

The findings are related back to the underlying literature. It is argued that an approach characterised by either genetic or environmental determinism fails to adequately describe the contingencies involved in the evocation and maintenance of challenging behaviours. The findings of the current thesis are discussed in the context of a developmental systems model (Moore, 2001), in which neither the influence of genetic nor environmental contributions can be fully understood without taking account of the other. This expanded model may hold important implications for the understanding of challenging behaviour.

The implications of the findings of the current thesis for the assessment, treatment and prevention of challenging behaviour are discussed. Some of the methodological limitations of the current thesis are noted, before ideas for future research are presented and final conclusions drawn.

The current thesis aimed to provide both a conceptual and empirical examination of the interaction between genetic and environmental factors in the development and maintenance of challenging behaviour displayed by children with intellectual and developmental disabilities. Findings from the previous four chapters are presented below.

Summary of Findings

In chapter two, a systematic literature review was conducted to identify areas in which future research was needed. The review functioned as a scoping exercise and focused on the concept of the motivating operation (MO; Laraway, Snycerski, Michael, & Poling, 2003; Michael, 1982, 1993) and its application to the functional analysis of challenging behaviour from 1998-2007. A number of areas for future research were identified. Of greatest relevance in the current context was the lack of empirical literature on interactions between basic behaviour-environment relations and genetic events. It has been argued that the MO may provide a useful means of describing such interactions (Kennedy, Caruso, & Thompson, 2001; McGill, 1999), however there has only been minimal empirical examination of this proposal or integration with existing models of the maintenance and development of challenging behaviour. The remainder of the thesis aimed to meet this need.

In chapter three, a model of the early development of self-injurious behaviour was presented. The model expanded existing operant models by incorporating gene-environment interactions (GxE) to account for why individuals with particular syndromes appear to be at a heightened risk of developing particular topographies of challenging behaviour. It was argued that genetic events may alter the development of the individual in a way that alters; i) the initial general movements of the child before they begin to form an operant response, ii) the discrimination of certain stimulus events, iii) the 'motivation' for some of the consequences that maintain challenging behaviour. A number of suggestions for future research that stem from this GxE model were proposed. Of particular relevance was the suggestion that future research be conducted that examines the potential motivative influence of genes upon behaviour-environment relations through the adoption of group-comparison designs.

To this end, chapter four presented a study in which within- and between-group differences in the function served by challenging behaviours displayed by children with FXS, SMS and a mixed etiology control group were examined using an indirect measure of behavioural function (the *QABF*).

There appeared to be some notable within-group differences for children with FXS. Children with FXS were significantly less likely to display attention-maintained than either escape- or tangible-maintained aggression or self-injurious behaviour, with a non-significant trend in the same direction for destructive behaviours. Children with FXS were also less likely to display pain-related behaviours than escape or tangible-maintained behaviours across all topographies. In contrast, the within-group pattern of results for children with SMS showed minimal differentiation[38]. Indeed, contrary to what had been predicted, children with SMS were no more likely to display attention-maintained challenging behaviour than any other function.

The between group comparison were generally supportive of these results. Children with FXS appeared to be less likely than either group to display attention-maintained challenging behaviours. Significant differences were found with the SMS group across all three topographies on this subscale and against the mixed etiology group for self-injurious behaviours and aggression. In contrast, children with SMS appeared to be more likely than either comparison group to display pain-related challenging behaviours. Significant differences were found against the FXS group for all three topographies on this subscale and against the mixed etiology group for self-injurious behaviours and aggression. No

[38] With the exception of automatically reinforced aggressive behaviours which, as expected, were significantly lower than all other functions for this topography in both the FXS and SMS groups.

between group differences were found for the tangible, automatic or demand subscales of the QABF.

To overcome some of the problems associated with indirect functional assessment methods, in chapter five, the function of challenging behaviours displayed by a group of eight children with FXS and six children with SMS was determined using experimental functional analysis methods. Each group was deemed representative of those who took part in the previous study.

There was notable individual variation in the occurrence of challenging behaviour. The pattern of results was, however, broadly consistent with those reported in the indirect study. Specifically, no child with FXS displayed any response class of challenging behaviour that appeared to be attention-maintained. In contrast, four children with SMS displayed a response class of challenging behaviour that was, at least in part, attention-maintained. Four of the six participants with SMS displayed challenging behaviours that were maintained by escape from demands and/or access to tangibles. This seems to support the findings of the previous study that children with SMS may not necessarily be any more likely to display attention-maintained behaviours than behaviour that serves other functions.

The text below provides a summary of the main findings from the two empirical studies.

1. *Within- and between-group comparisons showed that children with FXS were less likely to display attention-maintained challenging behaviour*

    - *Indirect study.* Children with FXS less likely to display attention- maintained behaviours than tangible- or escape-maintained challenging behaviours.
    - *Experimental study.* None of the eight children with FXS were found to display attention-maintained challenging behaviours, four displayed a response class

that was tangible-maintained and five displayed a response class that was escape maintained.

- *Indirect study.* Children with FXS less likely to display attention-maintained challenging behaviours than children with SMS or mixed etiology controls.
- *Experimental study.* In contrast to the FXS group, four of the six children with SMS displayed one response class of challenging behaviour that was at least in part attention-maintained.

2. *Children with SMS were no more likely to display attention-maintained challenging behaviours than challenging behaviours that served different social functions*

    - *Indirect study.* There were no within-group differences in the function served by challenging behaviour for children with SMS.
    - *Experimental study.* Four of the six children displayed at least one response class that was in part attention-maintained; three displayed at least one response class that was in part escape maintained, and two tangible-maintained.

3. *Children with SMS were more likely to display pain-related behaviours*

    - *Indirect study.* Children with SMS were more likely to be reported as displaying physical discomfort-related challenging behaviours than either children with FXS or mixed etiology.

These findings are now related back to the underlying literature and their implications for the understanding, assessment and treatment of challenging behaviour discussed.

Genetic Events as Motivating Operations

The term MO has been typically restricted to the description of variables that exert a momentary *value-* and *behaviour-altering* effect on operant behaviour. For example, sleep deprivation (Kennedy & Meyer, 1996; O'Reilly, 1995) and the absence of social contact (Berg et al., 2000; McComas, Thompson, & Johnson, 2003) have been shown to function as an EO for challenging behaviour displayed by individuals with intellectual and developmental disabilities. It has been suggested that the effect of certain genetic events may function as a type of MO, albeit whose effects are enduring rather than momentary (Kennedy et al., 2001; McGill, 1999; Oliver, 1993). For example, the value of food as a type of reinforcement appears to be enduringly established for individuals with PWS and the syndrome is characterized by hyperphagia. The genetic event that leads to PWS is associated with particular physiological changes that lead to an altered satiety response to food (see Kennedy et al., 2001). There appears to be many benefits to considering the effects of conditions such as PWS as a type of EO. Not only does this provide a conceptually systematic way of considering the behavioural function of such events (Langthorne, McGill, & O'Reilly, 2007; McGill, 1999) but it also has implications for models of the development and maintenance of challenging behaviours in individuals with particular syndromes as well as efforts for intervention and prevention. However, empirical support for this postulation has to date remained limited. The current thesis has provided some preliminary evidence to suggest that there may be both within- and between-group differences in behavioural function in individuals with different genetic syndrome. That is, aspects of certain syndromes may enduringly alter the value of certain events as types of reinforcement or punishment.

Children with FXS

For children with FXS the findings of the current thesis appear to be generally consistent with previous work to have examined behavioural function in this syndrome group. Two studies utilised indirect methods to examine behavioural function in FXS (Symons, Clark, Hatton, Skinner, & Bailey, 2003; Woodcock, Oliver, & Humphreys, 2009). Using an adapted version of the FAI, Symons et al reported that only 3% of boys with FXS displayed attention-maintained self-injurious behaviours, whereas 87% were reported to display such behaviour in response to changes in routine and 65% in response to task demands. The current study found similarly low levels of attention-maintained challenging behaviours occurring in children with FXS in comparison to escape- and tangible-maintained behaviours. In their study, which relied on a semi-structured interview similar in format to the FAI, Woodcock et al reported that over 75% of children with FXS were reported to display negative emotional behaviours and repetitive questioning following changes to routines or expectations. Boys with FXS were also reported to display significantly more anxiety-related self-injurious and repetitive behaviours following changes than were children with PWS. There is no data on the reliability or validity of the FAI, from which the measure of behavioural function adopted in both of these studies was derived. The use of the QABF in the current thesis to examine these relations therefore extends existing research on challenging behaviour in FXS; it is also unclear in the two cited studies as to whether the antecedent changes in routine relates to negatively reinforced (i.e., to escape from an aversive event) or positively reinforced behaviour (i.e., to access restricted tangibles). The use of the QABF in the current thesis was able to identify the maintaining variables involved in challenging behaviours displayed by children with FXS. In addition the between-group comparisons, which included a mixed etiology control group, served to extend these studies, which primarily involved within-group analysis.

Other studies have examined challenging behaviours in FXS using more direct behavioural measures. Hall, DeBernadis and Reiss (2006), for example, examined the behaviour of some 114 children with FXS across different social situations and found that challenging behaviours were more likely to occur across conditions in which there were high social or performance demands. Other studies have reported relatively high levels of behavioural problems occurring in similar situations that involve high social or performance demands (Hessl, Glaser, Dyer-Friedman, & Reiss, 2006; Lesniak-Karpiak, Mazzocco, & Ross, 2003). To the author's knowledge, no study to date has explicitly examined behavioural function in this group using experimental functional analysis methods. The findings from the functional analyses conducted in the current thesis appeared to suggest that children FXS were more likely to display challenging behaviours maintained by escape from demands or access to tangibles than behaviours that served other social functions, such as access to social attention. These findings were also consistent with the indirect findings from the study conducted in chapter four of the current thesis and those of other indirect studies (e.g., Symons et al., 2003). It was unclear from the Hall et al study whether challenging behaviours were evoked by the performance or social demands of the situations. The findings for the sample in the current study suggested that challenging behaviours were generally maintained by escape from academic demands rather than escape from attention.

In sum, children with FXS were found to be less likely to display attention-maintained behaviours than escape- or tangible-maintained challenging behaviours. The findings of the current thesis for children with FXS were generally consistent with those of previous studies but have extended the literature by utilizing validated modes of functional assessment (the QABF, experimental functional analysis) and between-group comparisons to address this question.

Children with SMS

The findings for children with SMS were not entirely consistent with the existing literature and as such require some further discussion. It has been suggested that children with SMS may be more likely to display challenging behaviours that are specifically maintained by social attention. For example, several clinical descriptions of the syndrome have noted the apparent 'insatiable need' for adult attention (Smith, Dykens, & Greenberg, 1998, p. 183). Other behavioural phenotype studies which used the Reiss Screen or the CBCL have found children with SMS tend to be score relatively highly on items relating to attention-seeking behaviours (Dykens, Finucane, & Gayley, 1997; Dykens & Smith, 1998). A within-group study by Sloneem (2005), which used the QABF as a measure of behavioural function, reported that children with SMS were more likely to display attention maintained verbal and physical aggression than other functions. No such differences were found for other behavioural topographies. Finally, Taylor and Oliver (2008) conducted a study which involved lag sequential analysis of the challenging behaviour. The study found that the pattern of challenging behaviour observed in four out of the five individuals with SMS in their sample was consistent with an attention-maintained hypothesis[39].

To some extent, the findings of the current thesis for children with SMS are consistent with the findings of other studies reported above. In the indirect study, children with SMS were found to be more likely to display attention-maintained self-injury, aggression and destructive behaviour than children with FXS and destructive behaviour than the mixed etiology group. In the experimental analysis study, four of the six children with SMS were found to display attention-maintained challenging behaviour. There were, however, also some notable discrepancies. The within-group analysis from the indirect study showed that children with SMS were no more likely to display attention-maintained behaviours than other social- and non-social functions. Likewise, in the experimental

[39] The reliance on descriptive functional assessment methods may have made it particularly difficult to detect the influence of variables that were delivered on a relatively lean schedule of reinforcement, however. It would seem particularly likely that such methods may be biased towards making a false positive result for attention-maintained behaviour. See chapter five for further discussion of the limitations of such methods.

analysis study, three children were shown to display challenging behaviour that was at least in part escape-maintained and two that was at least in part tangible-maintained. As such, it appears that rather than there being a unique association between SMS and attention-maintained behaviour, rather that children with SMS may be likely to display challenging behaviours that serve multiple functions, one of which may be attention.

Children with SMS were also more likely than either comparison group to be reported as displaying physical discomfort-related challenging behaviours. Although relations between health conditions associated with SMS and the occurrence of challenging behaviour have been described elsewhere (e.g., De Leersnyder, de Blois, Vekemans et al., 2001), this is the first study to document between-group differences in pain-related challenging behaviours using a measure of behavioural function. SMS is associated with a particular pattern of sleep disturbance (De Leersnyder, de Blois, Claustrat et al., 2001; De Leersnyder, de Blois, Vekemans et al., 2001) and peripheral neuropathy (Finucane, Dirrigl, & Simon, 2001) which can lead to severe pain. Health-related conditions, such as sleep disturbance or conditions associated with physical pain, have been found to influence challenging behaviour displayed by individuals with intellectual and developmental disabilities generally (see Kennedy & Becker, 2006; Langthorne et al., 2007; Symons, 2002). As such, it appears likely that such relations will also be present in genetic syndromes associated with particular health conditions, such as chronic sleep disturbance.

In sum, children with SMS were found to be likely to display challenging behaviours that served multiple functions, one of which was attention-maintained. The findings of the current thesis for children with SMS were partially consistent with those of previous studies. However, contrary to the suggestions of other studies (see L. Taylor & Oliver, 2008) no evidence of a unique association between SMS and attention-maintained behaviours was found. The use of a between-group comparison and experimental functional analysis methods extends the existing literature. In addition, children with SMS were found to be more likely than other groups to display pain-related behaviours. This is consistent with the findings of other studies, which were extended through the use of between group comparison and a recognised measure of behavioural function.

The within- and between group differences in behavioural function found in the current thesis provide some preliminary support for the suggestion that aspects of certain genetic conditions, such as FXS and SMS, can exert an enduring motivative influence on operant behaviour. Such GxE have potentially significant implications for theoretical models of challenging behaviour. In what follows, a developmental systems model, that incorporates GxE, and its implications for the study of challenging behaviour is presented.

GxE and a Developmental Systems Model

GxE

The findings of the current thesis suggest that gene-environment interactions (GxE) may hold important implications for the development of challenging behaviour in some individuals with intellectual and developmental disabilities. GxE refer to genotypic differences in susceptibility to an environmental pathogen for a given disorder (Cicchetti, 2007; Moffitt, Caspi, & Rutter, 2005; Rutter, Moffitt, & Caspi, 2006). For example, variations in the monoamine oxidase A (MAOA) gene have been repeatedly shown to moderate the effects of childhood maltreatment as an environmental 'pathogen' for the subsequent development of antisocial behaviour in the general population (Caspi et al., 2002; A. Taylor & Kim-Cohen, 2007). Similar relations may have an important effect on an individual's susceptibility or resilience to environmental causes of challenging behaviours displayed by people with intellectual and developmental disabilities (Hessl et al., 2008; May et al., in press).

It appears that challenging behaviour may be an optimum area for the further study of GxE[40]. However, little is known about the behavioural processes by which GxE influence the development of a particular disorder or psychopathology.

The concept of the *endophenotype* (Gottesman & Gould, 2003) refers to physical features, such as neurobiological changes, that uniquely vary with a given syndrome. The effects of genes on the developmental process involved in the emergence and subsequent maintenance of challenging behaviour must be via neuro-biological pathways (Moffitt et al., 2005). Such changes may represent the endophenotype of certain genetic syndromes (although variables at the cognitive level have also been invoked in this manner, see Woodcock, Oliver, & Humphreys, in press)[41]. Due to the level of experimental control that such studies afford, animal models may help in determining the effects of specific genetic events on neurobiological pathways. Such research may help explain why particular genetic syndromes are associated with particular behavioural functions.

Some recent work with animal models provides some illustrative examples of such work. A number of studies have used mouse models of stereotypy to investigate brain-behaviour-environment relations. Mark Lewis and colleagues in a series of studies examined brain-related changes associated with environmental enrichment in deer mice

[40] As Moffit, Caspi and Rutter (2005) state: "Wherever there is variation among humans psychological reactions to a major environmental pathogen for mental disorder, GxE must be expected to some degree (p.473).

[41] Woodcock, Oliver and Humphrys (in press) suggest a model to account for the repetitive questioning and temper outbursts associated with PWS that occur in response to changes in routine or expectations. In this model, the genetic event that gives rise to PWS is assumed to lead to changes in brain functioning that result in a cognitive endophenotype, a specific cognitive deficit in task switching. When high demands are placed on this cognitive ability (for example, by transitions or routine changes), it is argued that this elicits an emotional arousal response that is aversive for the individual and leads to temper outbursts or repetitive questioning.

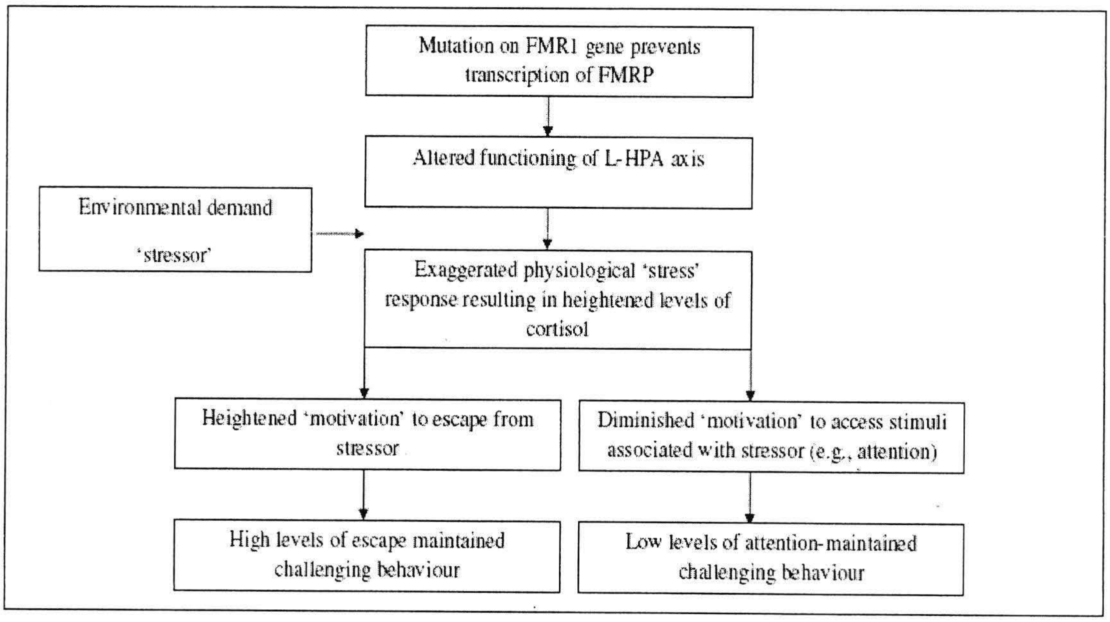
(see Lewis, Tanimura, Lee, & Bodfish, 2007; Turner & Lewis, 2003; Turner, Lewis, & King, 2003). Enrichment-related differences in brain circuitry, specifically in the motor cortex and basal ganglia regions, were found only in those deer mice that 'benefited' (i.e., as defined by the prevention or attenuation of stereotypical behaviours) from environmental enrichment. These enrichment effects were themselves associated with improved performance on learning tasks. Similar findings have been reported using a mouse model of FXS, Restivo et al (2005) found both behavioural and brain changes in FMR1-knockout mice following environmental enrichment. Mouse models have also shed some light on the neurobiological pathways involved in aggression. Couppis and Kennedy (2008) demonstrated that the opportunity to display aggression functioned as a positive reinforcement for a lever-pressing response in mice and that the reinforcing effectiveness of this consequence was itself modulated by alterations in D1 and D2 receptor activation in the nucleus accumbens. In a recent study, Couppis, Kennedy and Stanwood (2008) reported differences in brain circuitry in two mouse strains that differed in aggressive behaviour. Significant differences between aggressive and non-aggressive mice were found in the functioning of the mesocorticolimbic serotonergic and dopaminergic systems. Such studies provide one means by which the neurobiological pathways associated with particular genetic events and their relation to environmental events can be further examined.

It remains to be demonstrated how such factors influence susceptibility to the development of challenging behaviours that serve specific operant functions in individuals with FXS and SMS. However neuro-biological pathways have been implicated in the moderation of operant behaviour displayed by individuals with Prader-Willi syndrome. Holland, Whittington, and Hinton (2003) argue that the genetic abnormality underlying PWS leads to the impaired functioning of the hypothalamic pathways and the absence of metabolic and psychological changes that normally follow food intake. People with Prader-

Willi syndrome have been shown to have an abnormally high blood level of the hormone ghrelin, which may serve to enduringly heighten the reinforcing value of food (Del Parigi et al., 2002).

There may be neurobiological pathways involved in some of the relations described above for people with FXS and SMS. For example, FXS has been associated with the impaired functioning of the limbic-hypothalamic-pituitary-adrenal (L-HPA) axis, which plays an important role in the mediation of the human stress response. It has been suggested that the L-HPA axis may influence the occurrence of challenging behaviour in FXS, indeed positive correlations have been reported between levels of cortisol (an indicator of the functioning of the L-HPA axis) and parental report of behavioural problems (Hessl et al., 2002). Hypothetically, changes in brain circuitry that result from the mutation on the FMR1 gene that causes FXS, may lead to the altered functioning of the L-HPA axis. The onset of an environmental 'stressor', such as a demand, may lead to an exaggerated physiological stress response in children with FXS. This would be expected to enduringly heighten the child's 'motivation' to escape from such aversive stimuli and may explain the relatively high levels of negatively reinforced challenging behaviour observed in this group in the current thesis. One would also expect children with FXS to show a diminished 'motivation' for stimuli correlated with the onset of demands, such as attention, perhaps, accounting for the low levels of attention-maintained challenging behaviour observed for this group in the current thesis. Figure 6.1 provides a depiction of this hypothetical pathway between gene and behavioural function in FXS.

Figure 6.1. A GxE model of challenging behaviour in FXS



Developmental Systems Model

Evidence of the complex interplay between the biology of the individual and their environment suggests that the environmental and genetic determinism that has to date characterised the investigation of challenging behaviour may ultimately hamper the field and our ability to identify the determinants of such behaviour[42]. The findings above suggest that an expanded model is required to account for relations between variables at different levels of analysis, such as genes and environment, on the development of a response such as challenging behaviour.

It has been argued for some time that the nature-nurture debate is 'dead in the water' (S. M. Schneider, 2003). Rather than being caused by genes or environment, development is better understood as being driven by the 'coaction' of elements that form a

[42] As Schneider (2007, p. 93) notes: "Genetically determined" could be seen as useful shorthand in some cases, such as when a genetic feature…produces given effects across a large range of "normal" environments, just as, vice versa, "environmentally determined" could for an environmental feature that often produces given effects across a large range of "normal" genomes. But these simplifications can become problematic: They get overgeneralized, and the fact that they are simplifications can be forgotten."

single integrated system (Gottlieb, 1997; Moore, 2001; Oyama, Griffiths, & Gray, 2001). Both genes and environment must work together as part of this system to produce any aspect of any living thing (S. M. Schneider, 2007).

According to this developmental systems perspective an organism is comprised of a number of components that exist at several levels of analysis (genes, genes environment, cells environment, organ's environment and the macro environment). The environment in which the organism is embedded is simply viewed as another component of this system. Critically it is argued that each of the components of this system interacts with and affects the other. Thus, not only can variables at the same level of analysis influence the other (i.e., gene-gene relations, see Hessl et al., 2008) but that also variables at different levels of analysis may alter the influence of the other. These relations are completely bidirectional so that variables at a lower level of analysis may influence the effects of those that exist at higher levels and vice versa (i.e., not only can genes influence the effects of environmental variables, but also environment can moderate the influence of genes). Evidence of such bidirectional effects is relatively compelling. For example, Spira et al (2004) have demonstrated that cigarette smoke (an environmental pathogen) can lead to both reversible and irreversible changes in the expression of certain genes in human bronchial airway tissue. As has already been suggested genes may moderate the effects of environmental pathogens (Rutter et al., 2006). For example, the COMT gene has been shown to moderate the effects of heavy adolescent cannabis use as a pathogen for adult psychosis (Caspi et al., 2005).

Historically the application of behaviour analysis has been concerned with addressing only part of this developmental system (i.e., the influence of behaviour-environment relations). However, the radical behavioural philosophy that underpins applied behaviour analysis is consistent with the developmental systems model described above, stressing the importance of interactions between the individual and the context in which they are embedded (Chiesa, 1992; E. K. Morris, 1988; E. K. Morris & Midgley, 1990; Thompson, 2007). Schneider (2007) notes that behaviour analysis is entirely consistent with the developmental systems model, although its emphasis is on making the role of environmental factors and the behavioural principles therein explicit (p. 101). Within this approach, the question of whether a particular response is genetically or environmentally determined becomes redundant; rather the salient issue becomes what function each variable serves in relation to observable behaviour (Thompson, 2007).

One contribution behaviour analysis can make to the developmental systems model is to explicitly relate the influence of variables at varying levels of analysis (whether that be endogenous or exogenous to the organism) to underlying principles of behaviour. As Thompson (2007) notes, endogenous variables, such as genetic, hormonal and neurochemical influences, can alter the reinforcing value of certain behavioural consequences (see for example; Harvey et al., 2004; Kennedy, 2002a), can function as discriminative stimuli that set the occasion for a given response (e.g., Schuster & Brady, 1971), and can function as a reinforcing consequence for certain responses (e.g., Cataldo & Harris, 1982; Sandman, Spence, & Smith, 1999). Relating genetic or neurobiological variables to operant principles of behaviour will aid the delineation and understanding of the processes involved in GxE. As suggested in chapter three, such an enterprise may hold important implications for our understanding of challenging behaviours, such as self-injury.

In sum, the findings of the current thesis suggest that neither genetic nor environmental determinism provides an adequate model to account for challenging behaviour displayed by individuals with intellectual and developmental disability. GxE may play an important role in the development and subsequent maintenance of such behaviours. The developmental systems model, which is consistent with the tenets of radical behaviourism, provides a means with which both endogenous and exogenous variables can be brought to bear on the functional analysis of challenging behaviour.

Applied Implications

The finding that genetic events may exert a motivative influence on the consequences that maintain challenging behaviour also holds implications for the assessment, treatment and the prevention of challenging behaviours.

Perhaps the most significant applied implications of the current thesis lie in the design of prevention strategies for challenging behaviour in children with intellectual and developmental disabilities.

A number of studies suggest challenging behaviours typically emerge at a relatively young age in children with intellectual and developmental disabilities (e.g., Berkson, Tupa, & Sherman, 2001; Kurtz et al., 2003; Richman & Lindauer, 2005; M. J. Schneider, Bijam-Schulte, Janssen, & Stolk, 1996). Once established and in the absence of effective intervention such behaviours tend to be characterised by their chronicity (Murphy et al., 2005). It is already known that children with particular genetic syndromes, such as FXS or SMS, are at a heightened risk of developing particular forms of challenging behaviour, and as such are appropriate targets for secondary prevention efforts. It can frequently be difficult to identify the function of challenging behaviours in very young children, however (see Kurtz et al., 2003). Knowledge of the probable functions that challenging behaviours are likely to serve in young children with specific genetic conditions will aid efforts at secondary prevention, especially in cases where function is difficult to determine or challenging behaviour has not yet fully developed.

Such a preventative strategy would be based on those maintaining consequences for which 'susceptibility' is thought to exist. The current findings suggest that for children with FXS such early intervention work should be focused on ensuring that the child can *mand* to escape from aversive stimuli and access tangibles and that caregivers are responsive to these forms of communication. Efforts at reducing the aversive nature of demand presentation and on developing the child's ability to tolerate delay until reinforcement will also be important in this regard. For children with SMS the current findings suggest that the treatment of any health conditions, such as the sleep disturbance characteristic of the syndrome, should be done at as early a stage as is possible. It would seem important that additional secondary prevention efforts are focused on teaching the young child *mands* that serve multiple functions, one of which is to access attention.

The findings of the current thesis also have implications for the assessment and treatment of challenging behaviour in individuals where challenging behaviour is already established in their behavioural repertoire. Although no substitute for conducting a thorough functional assessment, it would be of benefit for clinicians to be aware that certain individuals are especially likely to display challenging behaviours that serve specific functions. For example, it would be useful to be aware that challenging behaviours displayed by an individual with SMS may be influenced by sleep disturbance. This would help prevent otherwise unexplained variability in the individual's challenging behaviour being attributed to unhelpful sources and help to guide intervention (Langthorne et al., 2007).

Limitations and Suggestions for Future Research

The current thesis has provided some preliminary evidence to suggest that there may be differences in the function served by challenging behaviours displayed by children with FXS and SMS. The two empirical studies in the current thesis aimed to complement one another and each was designed to compensate for some of the limitations of the other. Thus the experimental analogue study presented in chapter five was designed to overcome some of the problems associated with the indirect functional assessment methods used in the study presented in chapter four. Likewise, the larger sample size of the indirect study aimed to compensate for the low N necessitated by the use of experimental functional analysis methods. The correspondence between the findings of each study, despite the different methodologies used, is promising and augments the believability of the overall results.

Despite this there remain a number of limitations with the methods used in the current thesis which merit some consideration. A number of limitations with each empirical study have already been noted in the discussion section of each individual study. The following therefore focuses only on the more significant methodological constraints of each study. For discussion of less significant methodological issues the reader is referred to the relevant section of the discussion of each individual study in chapters four and five.

One of the most significant limitations affecting the indirect assessment study lies in the representativeness of the sample for each group. Convenience sampling was used to recruit participants for each group, with the majority of participants recruited via parental support groups. It may be that these participants were not sufficiently representative of the underlying population for each syndrome group.

An issue affecting both studies is that the author and lead researcher was not blind as to the genetic status of each participant and the aims of the study. Although every attempt was made to mitigate against the effect of this, for example by closely following the study protocol, it is possible that this influenced the responding of participants in both studies.

Chapter two provided a systematic review of the concept of the MO and its application to the functional analysis of challenging behaviour. The review contained reference to a number of potential threats to the internal validity of analogue functional analysis methods. For example, recent research on the behaviour-altering effect of the MO (O'Reilly, Edrisinha, Sigafoos, Lancioni, & Andrews, 2006; O'Reilly et al., 2007; O'Reilly, Sigafoos et al., 2006) suggests that if left uncontrolled MOs, such as the deprivation of attention or tangibles, can evoke challenging behaviour even in conditions where the relevant discriminative stimuli and reinforcing consequences for the behaviour are absent.

Thus, challenging behaviour evoked by the deprivation of tangibles could occur within the demand or attention condition of the functional analysis[43]. Whilst such confounds are not beyond experimental control, by making pre-session manipulations for example, they do constitute a threat to the internal validity of experimental functional analysis generally. In the current study no efforts were used to control such influences other than those typically used when conducting such analyses (e.g., randomized order of experimental conditions). It is possible therefore that there were uncontrolled confounds that influenced the responding of individual participants. Such threats apply to all experimental functional analyses and with only a few exceptions (see Volkert, Lerman, Call, & Trosclair-Lasserre, 2009) tend not to be explicitly addressed in advance of conducting a functional analysis. Some consensus is required within the field for feasible means of dealing with such potential confounds if the internal validity of analogue assessment methodologies is not to be undermined.

Some concerns remain with the external validity of the experimental functional analysis methods used in the current thesis. For a number of participants in both groups the findings of the experimental analyses did not correspond to the situations reported by caregivers as evoking challenging behaviour. For example, Abe was reported by care-givers to display escape-maintained challenging behaviours in response to academic demands and this was observed firsthand by the author. However, one session aside, this did not occur within the confines of the experimental functional analysis. Although all analyses were conducted in the child's natural settings, it may have been that the use of the author, with whom the child had limited learning history, as the 'therapist' in the functional analysis may have resulted in such discrepancies. Differences in the results of functional

[43] Other potential threats include the influence of toy preference and type of attention provided on challenging behaviour in the attention condition, the means of task presentation in the demand condition, the influence of other extraneous variables, such as health factors.

analyses dependent on whom is conducting the assessment have been reported in the literature (e.g., Ringdahl & Sellers, 2000) and it may be that the use of the child's caregivers to run the functional analysis may have provided a more externally valid methodology. Although beyond the scope of the current thesis, due to the additional practical resources required to train caregivers to implement such assessments, future research should consider means of increasing the external validity the experimental functional analysis methods used to assess between-group differences in behavioural function.

The experimental functional analysis study did not have sufficient N to enable meaningful statistical comparison between the groups. Thus it is unclear as to whether the differences observed in the study were similar to what would be expected as a result of normal sampling error. Whilst the results of the indirect study mitigate this concern to some extent; given the status of experimental functional analysis as the 'gold standard' of functional assessment methodologies (Hanley, Iwata, & McCord, 2003) it seems critical that future research be conducted that adopts experimental functional analysis methods and uses sufficient N to facilitate meaningful statistical comparison. Given the resource intensive nature of experimental functional analyses and the low incidence of individuals with genetic syndromes such as SMS, large-scale, collaborative, multi-site studies may be required to meet the practical demands of such work. The use of existing comparative data on the prevalence of different behavioural functions in people with intellectual and developmental disabilities generally (e.g., Derby et al., 1992; Hanley et al., 2003; Iwata et al., 1994) will also provide a meaningful comparison against which experimental groups can be compared. The current thesis suggests that such an endeavor may be worth serious consideration.

As well as replicating the existing studies with larger N, it would be of interest to test the gereralizability of these findings with different syndrome groups. The author is aware of some ongoing research that is examining the function of challenging behaviour in children with Angelman, Cri Du Chat and Cornelia De Lange syndromes (Tunnicliffe et al., 2008), however research is still needed in this area. Such relations, as discussed in previous chapters may be relatively commonplace and it would seem important that both within- and between-syndrome similarities and differences in behavioural function are properly delineated. Such efforts are likely to require collaborative, multi-site efforts.

Future research should elucidate on the proximal effects of genes that may give rise to the differences in behavioural function reported in the current study. The concept of the *endophenotype* (Gottesman & Gould, 2003) has received some recent attention in this regard in relation to challenging behaviour in PWS (see Woodcock et al., 2009, in press). It would be of interest to begin to examine the endophenotype associated with FXS and SMS and specifically to examine how this relates to the differences in behavioural function observed in the current study. This strategy would involve the analysis of variables at a different level of analysis to those examined in the current thesis; however a similar strategy has proved useful in the investigation of the neural underpinnings that underlie the relation between sleep and escape-maintained behaviour (see Harvey et al., 2004). Such studies could potentially increase the options for intervention beyond solely environmental manipulation.

Finally, a proposal made in chapter three and reiterated here is that future research be conducted that examines differences in challenging behaviour as it develops in young children with specific genetic syndromes. Some data does already exist in this regard for children with Lesch-Nyhan syndrome (Hall, Oliver, & Murphy, 2001), however it would be of interest to empirically examine whether the 'motivative' effects of certain genetic events accelerates the mutual reinforcement processes already known to play a causal role in the development of challenging behaviour in children with intellectual and developmental disabilities (Kennedy, 2002b; Oliver, Hall, & Murphy, 2005; Richman, 2008), as suggested in chapter three of the current thesis and by other researchers in the field (e.g., L. Taylor & Oliver, 2008).

Final Conclusions

The current thesis has provided both a conceptual and empirical analysis of GxE as applied to challenging behaviours displayed by children with intellectual and developmental disabilities. It appears that one role of genetic events may be to exert a motivative influence over some of the social and non-social consequences that maintain challenging behaviour. Preliminary evidence of the influence of such relations on challenging behaviour was provided for children with FXS and SMS. Children with FXS were less likely to display attention-maintained challenging behaviours. Children with SMS were found to display challenging behaviours that served multiple functions (one of which may have been attention-maintained) and were more likely than other groups to display pain-related challenging behaviours. The behaviour analytic concept of the MO may help to describe some of the *value-* and *behaviour-altering* effects of such events. Despite some of the limitations of the current thesis, it appears that these findings have considerable implications for the assessment, treatment and prevention of challenging behaviour as well as for models of the initial development and subsequent maintenance of such behaviours.

The probable relevance of such relations has been noted since the early infancy of behaviour analysis. As Sidney W Bijou stated:

> *Psychological development consists of progressive changes in interactions between the individual, as a total functioning biological system, and the environmental events (Bijou, 1966, p. 2).*

Further elucidation of GxE and the role they play in challenging behaviours displayed by people with intellectual and developmental disabilities will help to bring the application of operant science more in line with its theoretical foundations.

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Appendices

Appendix 1

MO Studies for Attention-Maintained Challenging Behaviour

Studies	N	Age	Challenging behaviour (s)	Independent Variable	Design	Findings
Piazza, Hanley et al (1998)	2	4-8	Aggression; disruption; SIB	Effects of break from demands	Multi-element embedded in reversal design (Tom); multi-element (Jerry)	Break from demands evoked attention-maintained problem behaviour
Northup et al (1999)	4	7-8	Inappropriate vocalizations; out of seat; playing with objects; off-task behaviours	Effects of methylphenidate (MPH)	Multielement embedded in super ordinate multielement design	MPH influenced problem behaviour of three participants
O'Reilly (1999)	1	20	Yelling; SIB	Effects of pre session manipulation	Reversal (ABAB)	Pre session deprivation of attention led to higher rates of challenging behaviour
O'Reilly et al (1999)	2	8-31	Aggression; SIB	Effects of pre-session manipulation	Multi-element embedded in reversal design	Pre session exposure to diverted attention condition led to higher rates of challenging behaviour
Roane et al (1999)	5	5-21	Aggression; inappropriate vocalization; SIB; disruptive behaviour	Effects of within-session manipulation	Multielement design	Greater likelihood of problem behaviour when maintaining variable (attention) absent
Berg et al (2000)	3	1-4	SIB; pulling and chewing medical equipment;	Effects of pre session manipulation	Multi-element design	Challenging behaviour influenced by manipulations made to pre-session attention

Fisher et al (2000)	1	17	SIB; aggression; disruption	Effects of tangibles	Multi-element design	Attention-maintained problem behaviour reduced to zero levels in conditions where high preferred tangibles present. Low preferred tangibles had no influence
O'Reilly, Lancioni et al (2000)	2	9-22	Pushing; pinching; SIB; property destruction	Effects of modified attention condition	Multi-element design	Diverted attention condition evoked problem behaviour. Reduced when attention provided non-contingently.
Ringdahl & Sellers (2000)	3	5-13	SIB; aggression	Effects of different individuals	Multi-element embedded in reversal design	Attention-maintained problem behaviour of two individuals only apparent when caregiver ran FA
Worsdell et al (2000)	6	29-52	SIB	Effects of within-session manipulation	Multi-element design	Consistently high rates of challenging behaviour only evoked in EO sessions with reinforcement contingency
LeBlanc et al (2001)	1	11	Aggression	Effects of different individuals	Multi-element embedded in reversal design	Both gender of therapist and type of attention provided influenced challenging behaviour
Ringdahl et al (2002)	2	5-41	Aggression; switch activation	Effects of tangibles	Multi-element embedded in	Availability of toys in attention

					reversal design	condition reduced attention-maintained challenging behaviour
McComas et al (2003)	5	8-12	Throwing materials; hitting; spitting; property destruction; pinching; scratching; kicking	Effects of pre-session manipulation	Multi-element	Pre-session attention influenced attention- but not escape-maintained problem behaviour
English & Anderson (2004)	4	5-16	Aggression; disruption; SIB	Effects of different individuals	Multi-element embedded in reversal design	Presence or absence of caregivers influenced challenging behaviour
Call et al (2005)	2	2-17	Aggression; destruction	Effects of modified attention condition	Multi-element	Higher rates in combined conditions (i.e., demand & diverted attention/cont att)
Dicesare et al (2005)	1	18	Disruption	Effects of MPH	Multi-element embedded in reversal design	Higher rates of attention-maintained behaviour when no MPH; low levels across all conditions when MPH present
Harding et al (2005)	2	1-5	SIB	Effects of location	Multi-element (Phase 1/3); reversal (Phase 2)	Location change allowed identification of social function for previously undifferentiated challenging behaviour
O'Reilly, Edrisinha et al (2006)	1	20	Bizarre speech; elopement	Behaviour-altering effect	Multi-element design	High rates of attention-maintained behaviour following pre-session deprivation in both reinforcement and extinction conditions.

O'Reilly, Sigafoos et al	2	14-20	SIB; aggression; elopement hair eating; self-touching	Behaviour-altering effect	Multi-element design	High rates of challenging behaviour (2006) following pre-session deprivation in both reinforcement and extinction conditions.
Roantree & Kennedy (2006)	1	10	Stereotypy	Effects of pre-session manipulation	Multi-element embedded in reversal design	Attention function following pre-session attention and undifferentiated following pre-session alone
O'Reilly, Edrisinha et al (2007)	1	20	Bizarre speech; elopement	Behaviour-altering effect	Multi-element	Higher rates of problem behaviour in both alone and extinction conditions following pre-session deprivation of attention

Appendix 2 *MO Studies for Tangible-Maintained Challenging Behaviour*

Studies	N	Age	Target behaviour (s)	Independent Variable	Design	Findings
Fisher et al (1998)	2	13-14	Aggression; dangerous behaviour; property destruction	Effects of interruption to free operant behaviour	Multi-element design (Phases 3/4; reversal (ABAB) (Phase 5)	Challenging behaviour occasioned by requests that interfered with ongoing activity (e.g., watching game shows and looking out of window)
Adelinis & Hagopian (1999)	1	27	Aggression	Effects of interruption to free operant behaviour	Reversal (ABAB)	High rates of challenging behaviour occasioned by requests that interfered with ongoing activity (e.g., lying of the floor). Not occasioned by symmetrical 'do' requests.
Roane et al (1999)	5	5-21	Aggression; inappropriate vocalization; SIB; disruptive behaviour	Effects of within-session manipulation	Multielement design	More challenging behaviour occurred when tangibles (e.g., toys) absent than when present
Murphy et al (2000)	1	38	Aggression; stereotypical behaviour	Effects of interruption to free operant behaviour	Multi-element design	Challenging behaviour more likely to occur in conditions in which prevented from engaging in free operant response (e.g., flushing objects down toilet).
Worsdell et al (2000)	6	29-52	SIB	Effects of within-session manipulation	Multi-element design	Consistently high rates of

						challenging behaviour when tangibles (e.g., food) absent and reinforcement contingency present
Mueller et al (2001)	1	8	Aggression	Effects of stimulus preference	Multi-element embedded in reversal design (ABCA)	Restricting high preferred items (e.g., books) evoked highest rates of challenging behaviour, restricting less preferred (e.g., stuffed animal) associated with moderate rates of aggression.
Ray & Watson (2001)	3	4-5	Aggression; out-of-seat behaviour	Effects of pre-session influences	Multi-element design	Undifferentiated FA revealed tangible function when examined correlation with prior events
Asmus et al (2003)	1	7	Disruptive behaviour; stereotypical behaviour	Effects of reinforcement for alternative response classes	Multi-element in AB design	Absence of reinforcement for stereotypy increased rate of destructive behaviour maintained by tangibles(toy train)
McLaughlin et al (2003)	1	5	Aggression; tantruming; destruction	Effects of stimulus preference	Reversal design	Challenging behaviour only occurred in conditions that led to active play
Fritz et al (2004)	2	12-19	Aggression; disruption; Dropping	Effects of interruption to free operant behaviour	multiple-baseline; reversal	Termination of preferred activity (e.g., walking) functioned as EO for tangible- maintained problem

						behaviour in the context of demands
McAdam et al (2004)	1	18	Aggression; disruption	Effects of different individuals	ABC design	Tangible (access to leisure items)- and escape-maintained behaviour in conditions run by caregivers none in conditions run by therapist
Harding et al (2005)	2	1-5	SIB	Effects of location	Multi-element (Phase 1/3); reversal (Phase 2)	Location change allowed identification of social function for previously undifferentiated challenging behaviour
O'Reilly, Sigafoos et al (2006)	2	14-20	SIB; aggression; elopement hair eating; self-touching	Behaviour-altering effect	Multi-element design	High rates of challenging behaviour following pre-session deprivation in both reinforcement and extinction conditions for challenging behaviour maintained by edibles
Wilder et al (2006)	2	3	Aggression; tantrums	Effects of interruption to free operant behaviour	Multi-element	Challenging behaviour of one child occurred exclusively when asked to terminate a preferred a activity (e.g., watching a video, playing with toy cars)
Hagopian et al (2007)	3	6-12	SIB ; aggression ; disruption	Effects of interruption to free	Pairwise design (Perry/Kelly);	High rates of challenging

				operant behaviour	reversal design (Maxwell)	behaviour following both 'do' and 'don't' requests (e.g., playing with toys)
O'Reilly, Edrisinha et al (2007b)	1	14	SIB; aggression	Behaviour-altering effect	Multi-element design	High rates of challenging behaviour following pre-session conditions in which no access to edibles

Appendix 3

MO Studies for Negatively Reinforced Challenging Behaviour

Studies	N	Age	Challenging behaviour (s)	Independent Variable	Design	Findings
Asmus et al (1999)	3	3-5	SIB, aggression, destruction disruption	Effects of demand stimulus parameters	Multi-element	For two participants presentation of unfamiliar tasks reduced challenging behaviour.
Garcia & Smith (1999)	2	29-42	SIB	Effects of naltrexone	Multi-element embedded in double blind, placebo controlled reversal design	Escape-maintained head slapping of one participant reduced in naltrexone conditions
Roane et al (1999)	5	5-21	Aggression; inappropriate vocalization; SIB; disruptive behaviour	Effects of within-session manipulations	Multielement design	More challenging behaviour occurred when demand present
McComas et al (2000)	3	8-9	Destructive behaviour	Effects of demand stimulus parameters	Multi-element	Presence of instructional strategy, opportunities for choice-making, and task repetition shown to influence challenging behaviour
O'Reilly et al (2000)	1	5	Aggression, destructive behâviour, pain behaviours	Effects of demand stimulus parameters	Multi-element embedded in reversal design	All background noise conditions evoked pain related behaviours.

						Challenging behaviour only occurred in conditions where both demand and noise were present
O'Reilly & Lancioni (2000)	1	4	SIB; aggression	Effects of sleep deprivation	Reversal design	Correlation between sleep deprivation and escape-maintained SIB but not aggression
Ducharme & Rushford (2001)	1	10	Compliance	Effects of pre-session manipulation	Reversal design	Child compliance increased following pre-session play conditions
Hagopian et al (2001)	1	6	SIB; aggression; disruption; Spitting	Effects of other stimulus events	Multi-element	Challenging behaviour evoked by social attention
McCord, Iwata et al (2001)	7	25-44	SIB; aggression; tantrum; property destruction	Effects of other stimulus events	Multi-element	Challenging behaviour evoked by onset of noise.
McCord, Thompson, et al (2001)	2	27-38	SIB	Effects of other stimulus events	Multi-element	Challenging behaviour of two individuals evoked by onset of transitions that involved change in location. Some demands evoked challenging behaviour independent of location
Moore et al (2001)	4	4-9	Aggression; destructive behaviour; disruptive behaviour;	Effects of other stimulus events	Multi-element embedded in reversal design	Challenging behaviour of two participants evoked by high levels of attention not task

						demands.
Oliver et al (2001)	1	14	Aggression	Effects of other stimulus events	Alternating treatment	Challenging behaviour occurred in high attention conditions only when therapist in close proximity . Extinguished over extended assessment periods.
Carey & Halle (2002)	1	12	SIB	Effects of combined antecedents	Multi-element design	Challenging behaviour occurring in demand contexts reduced when music available
Romaniuk et al (2002)	7	7-10	Aggression; disruptive behaviour	Effects of demand stimulus parameters	Reversal design	Choice making reduced escape-maintained behaviour occurring in demand contexts but no influence on attention-maintained behaviour
Carr, Magito-McLaughlin et al (2003)	8	29-48	SIB; aggression; property destruction	Effects of pre-session influences	Reversal design	Correlation between 'bad mood' rating and occurrence of problem behaviour only in demand contexts
Carr, Smith et al (2003)	4	26-31	SIB; aggression; property Destruction; tantrums	Effects of menses	Reversal design	Higher levels of problem behaviour in demand contexts when menses present
Ebanks & Fisher (2003)	1	9	SIB; aggression; property destruction	Effects of demand stimulus parameters	Reversal design	Providing corrective prompt after task evoked challenging

						behaviour providing pre-task prompt reduced challenging behaviour
Moore & Edwards (2003)	4	7-17	Disruptive behaviour; property destruction	Effects of other stimulus events	Multi-element	Idiosyncratic influences across individuals. Teacher attention functioned as aversive stimuli for two individuals; onset of tasks for remaining participants
Borrero et al (2004)	1	13	Aggression	Effects of demand stimulus parameters	Reversal design	Challenging behaviour in demand contexts occurred more frequently when used abrasive rather than pleasant prompting style
Call et al (2004)	6	4-8	Aggression; destruction; tantrums; off-task behaviour	Effects of combined antecedents	Multi-element	Attention appeared to attenuate aversiveness of demands for three individuals. Altering rate and/or difficulty of demands reduced challenging behaviour for three individuals
McAdam et al (2004)	1	18	Aggression; disruption	Effects of different individuals	ABC design	Tangible- and escape-maintained behaviour in conditions run by caregivers none in conditions run by therapist
Call et al (2005)	2	2-17	Aggression; destruction	Effects of combined antecedents	Multi-element	Higher rates in combined

						antecedent conditions (i.e., demand & restricted tangibles with contingent escape)
Long et al (2005)	3	5-19	SIB; aggression; disruption	Effects of demand stimulus parameters	Reversal	Providing preferred stimuli reduced challenging behaviour that occurred in the context of hygiene-related demands
Magito-McLaughlin & Carr (2005)	3	28-39	SIB; aggression	Effects of different individuals	Reversal (study 1); multiple-baseline (study 2)	Higher rate of challenging behaviour in demand contexts when presented by poor 'rapport' dyad. Challenging behaviour reduced and task completion improved following 'rapport' intervention
Patel et al (2005)	3	3-4	Food packing; food acceptance; inappropriate behaviour; expulsion; vomiting	Effects of other stimulus events	Reversal design	Higher textured food associated with higher levels of packing and less food intake than lower textured food
Peyton et al (2005)	1	10	Disruptive behaviour	Effects of demand stimulus parameters	Multi-element	Escape-maintained problem behaviour evoked not by task per se but by directive prompts
Buckley & Newchok (2006)	1	7	Ear covering; screaming	Effects of other stimulus events	Multi-element	Source of music shown to be critical variable in evoking challenging behaviour
Crockett & Hagopian (2006)	1	19	SIB; aggression; property	Effects of demand stimulus parameters	Multiple-baseline	Higher rates of problem

			destruction; task completion			behaviour and fewer task completion when used three-step prompting procedure
Kelley et al (2006)	1	11	SIB; aggression; disruption;	Effects of amphetamine	Reversal design	Allocated responses to compliance in drug conditions and to problem behaviour in placebo conditions
Boelter et al (2007)	3	3-6	Aggression; destructive behaviour; non-compliance	Effects of demand stimulus parameters	Multi-element design	Idiosyncratic pattern of results. One-step directives set the occasion for accurate responding and other aspects of the task (e.g., preference, 3-step prompts) evoked challenging behaviour
Potoczak et al (2007)	4	7-17	Destructive behaviour; non-compliance; out-of-seat behaviour	Effects of within-session manipulation	Test-control multi-element	ABC method (EO with reinforcement contingency) resulted in differentiated pattern of responding for all 4 participants. AB method (EO/no reinforcement contingency) more ambiguous

Appendix 4

MO Studies for Automatically Reinforced Challenging Behaviour

Studies	N	Age	Challenging behaviour (s)	Independent Variable	Design	Findings
Fisher et al (1998)	2	7-15	Property destruction; stereotypical behaviour	Effects of matched stimuli V competing stimuli	ABAB (Exp 1/3); multi-element design (Exp 2)	Lower levels of property destruction and stereotypy when given access to matched than unmatched toy.
Piazza et al (1998)	3	4-17	Pica	Effects of matched stimuli V competing stimuli	Multi-element embedded in reversal design (study 3, study 4 Mary only); multi-element (study 4, Brandy)	Lower levels of pica when had access to matched as opposed to unmatched stimuli, subsequent analysis revealed texture was critical variable
Roscoe et al (1998)	3	20-35	SIB	Effects of competing stimuli	Multiple-baseline with embedded multi-element design	Competing stimuli reduced SIB
Thompson et al (1998)	1	7	Chin-grinding; aggression	Effects of matched stimuli	Multi-element design	Matched stimulation *with* response blocking led to greater reductions in challenging behaviour.

Garcia and Smith (1999)	2	29-42	SIB	Effects of naltrexone	Multi-element embedded in double blind, placebo controlled reversal design	Reductions in automatically reinforced SIB of one participant
Goh et al (1999)	4	40-49	Cigarette pica	Effects of matched stimuli	Multiple-baseline	Providing matched stimuli led to reductions in pica but did not maintain.
Rapp et al (1999)	1	19	Hair pulling; hair manipulation	Effects of matched stimuli	Multi-element design	Lower rates of hair pulling when given non-contingent access to previously pulled out hair
DeLeon et al (2000)	1	11	SIB	Parameters of competing stimuli	Multi-element embedded in a reversal design	Lower levels of SIB when multiple toys available. Within-session analysis revealed that in single-set condition toy play decreased and SIB increased over the course of the session
Friman (2000)	1	3	Thumb sucking	Effects of presence of specific stimuli	Multiple-baseline with reversal design	Thumb sucking only occurred in presence of cloth.
Patel et al (2000)	2	10-30	Head-hitting; rapid tongue movements	Effects of matched stimuli	Multi-element design (Phase 2); Multi-element embedded within a ABAB design (Phase 4)	Providing access to tactile and auditory stimuli reduced problem behaviour

Piazza et al (2000)	3	6-17	Dangerous behaviour; hand-Mouthing; saliva manipulation	Effects of matched stimuli V competing stimuli	Multi-element (Betsy); multi-element embedded in reversal design (Brad/Tyrone)	Challenging behaviour reduced when given access to matched stimuli a but not when given access to unmatched stimuli
Rapp et al (2000)	1	4	Hair manipulation; object manipulation	Effects of matched stimuli	Multi-element design	Low levels of hair-pulling when given access to matched source of stimulation.
Van Camp et al (2000)	2	5-8	Hand-biting; hand flapping; toy play	Effects of presence of specific stimuli	Multi-element design and ABAB reversal	Presence of ball and social interaction occasioned problem behaviour of two individuals
Wilder et al (2000)	1	30	Head rocking	Effects of competing stimuli	Multi-element design; (Phase 2), multi-element embedded in reversal (Phase 3)	Lower levels of head rocking when given massage to shoulders and neck. Providing enriched environment (EE) with vibrating pen reduced head rocking but EE without pen did not
Cuvo et al (2001)	4	44-65	Stereotypical behaviours	Effects of competing stimuli	Multi-element design	Differences in stereotypical behaviour across settings with differing levels of stimulation
Healey et al (2001)	1	21	SIB	Effects of competing stimuli	Multiple-baseline design with embedded reversal	Lower levels of SIB when competing stimuli available
Britton et al (2002)	3	8-28	Stereotypical behaviours	Effects of matched stimuli	Multi-element design; reversal	Stereotypy reduced when had access to matched sources of

						stimulation with prompting
Carr et al (2002)	1	7	Object mouthing	Effects of matched stimuli V competing stimuli	Reversal with embedded multi-element component	Challenging behaviour reduced only when combined NCR with response blocking. No difference between matched and unmatched stimulation
Lindberg et al (2003)	3	30-43	SIB	Parameters of competing stimuli	Reversal design	Brief noncontingent access to single preferred stimuli reduced SIB for all 3 individuals. When extended sessions led to increase in SIB and reduction in object manipulation for 2 participants until varied reinforcers available.
O'Reilly et al (2003)	1	27	Attempts at SIB	Effects of competing stimuli	Multi-element design (Phase 2 and 3); Reversal design (Phase 3)	Attempts at SIB reduced when noncontingent access to vibrating stimuli provided but not when given access to alternative forms of stimulation.
Roane et al (2003)	1	8	Object mouthing	Effects of competing stimuli	Multiple-baseline	Competing stimuli reduced challenging behaviour
Simmons (2003)	1	48	SIB	Effects of matched stimuli	Multiple-schedule design with reversal	Evaluated effects of providing noncontingent access to food on SIB. Measures of SIB post-intervention were lower than pre

Tang et al (2003)	6	4-17	Stereotypical behaviour	Effects of matched stimuli	Multi-element	Lower levels of stereotypy in specific sensory masking conditions for four participants. Providing matched stimulation reduced stereotypy of three participants.
Carter et al (2004)	1	8	screaming; hand-mouthing	Effects of presence of specific stimuli	Multi-element embedded in reversal design	Challenging behaviour occurred only in those conditions in which toys present
Rapp (2004)	1	10	object twirling	Effects of response deprivation/satiation and presence of specific stimuli	Multi-element embedded in reversal design	Stereotypy lower in second session of the day suggesting prior access to reinforcer acted as AO. Higher levels of object twirling when music played
Rapp et al (04)	5	5-14	Stereotypy	Effects of response deprivation/satiation and competing stimuli	ABAB design	For 3 participants, highest level of challenging behaviour occurred after response restriction. Providing stimulation matched to sensory products of behaviour reduced stereotypy of one participant.
Ahearn et al (2005)	2	11-13	Stereotypy	Effects of matched stimuli V competing stimuli and parameters of competing stimuli	Multi-element	Non-matched but highly preferred stimuli led to greater reductions in stereotypy of two participants

Carter (2005)	1	4	SIB	Effect of sinus infection and weighted vest	Multi-element	Higher levels of SIB when sinus infection present than absent. Weighted vest had no influence on SIB.
Higbee et al (2005)	1	12	Stereotypy	Effects of matched stimuli V competing stimuli	Reversal	Stereotypy reduced when given access to matched form of stimulation, no reductions when given access to highly preferred unmatched stimuli
Long et al (2005)	3	5-19	SIB; aggression; disruption	Effects of competing stimuli	Reversal	Providing competing stimuli reduced automatically reinforced challenging behaviour
Rapp (2005)	3	9-11	Stereotypy	Effects of presence of specific stimuli	Multiple probe design (Exp 1; Carl & Ryan; Exp 2/3 Ryan only) design. Reversal (Exp 1; Andy; Exp 2/3 Carl & Andy)	Audio and visual stimulation exerted an influence on various topographies of stereotypy
Sidener et al (2005)	2	6	Stereotypy	Effects of matched stimuli	Multiple baseline design with embedded reversal	Consequence-based intervention had no impact on problem behaviour, but stereotypy reduced following NCR with matched stimuli
Taylor et al (2005)	1	4	Stereotypy	Effects of matched stimuli V competing stimuli	Multi-element design; (Phase 2) concurrent operant choice procedure (Phase 3)	Less vocal stereotypy when given access to auditory toys.

Rapp (2006)	1	9	Stereotypy	Effects of response deprivation/satiation and matched stimuli	Multiple schedules	Higher levels of stereotypy following response restriction; lower levels of stereotypy following access to matched stimuli
Kenzer & Wallace (2007)	1	59	Rumination	Effects of competing stimuli	Reversal design	Food satiation reduced problem behaviour
Rapp (2007)	2	9	Stereotypy	Effects of matched stimuli V competing stimuli	Reversal design; (Exp 1) Multiple-schedule and reversal (Exp 2)	Providing matched stimulation led to greater reductions in stereotypy than unmatched. Stereotypy of both participants remained below pre-intervention levels following the removal of auditory stimulation

Appendix 5. Inter-rater reliability data.

Participant	Response class	Reliability Total	Reliability Occurrence	Reliability Non-occurrence	Kappa
Abe	Finger-biting	99%	92.1%	98.9%	.95
	Other CB	99.3%	87.5%	99.3%	.93
Greg	Head-hitting	100%	100%	100%	1.0
	Other CB	99.1%	95.4%	98.9%	.97
Jacob	CB	99.2%	87.5%	99.1%	.93
Luke	CB	97.8%	90.9%	97.1%	.94
Theo	CB	99.3%	89.5%	99.2%	.94
John	Finger-biting	99.6%	87.5%	99.6%	.93
Calum	CB	99.7%	97.9%	99.6%	.99
Richard	CB	99%	96%	99%	.97
Sean	Self-biting	98.8%	83.3%	98.6%	.90
	Other CB	94.1%	85.1%	90.4%	.87
Matt	Object banging	98.1%	75.9%	97.9%	.85
	Other CB	98.3%	87.5%	98.1%	.92
Angus	Finger-biting/object mouthing	99.6%	88.9%	99.6%	.94
	Other CB	98.3%	81.8%	98.1%	.89
Patrick	Head tapping	99.4%	85.7%	99.4%	.92
Shelley	Lying on floor	98.3%	95.5%	97.4%	.97
	Other CB	98.3%	80.9%	91.2%	.89
Katie	Stereotypical behaviour	99.6%	95%	99.5%	.97
	Other CB	100%	100%	100%	1.00