VOLUME I

RESEARCH COMPONENT

Antecedent Influences on Negatively Reinforced Behaviour. An Examination of Person-Environment Interplay.

by

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Abstract

This thesis comprising both research and clinical volumes is submitted in partial fulfillment of the degree of Doctor of Clinical Psychology (DClinPsy) at the University of Birmingham.

Volume I contains a literature review, an empirical paper and a public domain briefing paper. The literature review provides a systematic review of the literature examining the role of *motivating operations* for problem behaviour maintained by negative reinforcement. It provides a critical review of the existing evidence together with suggestions for future research. A model of problem behaviour emphasizing the role of person-environment interactions is provided. The empirical paper describes an empirical examination of such interactions within fragile X syndrome. The influence of experimental manipulations made to the degree of eye contact on gaze avoidance and indices of arousal in four boys with a diagnosis of fragile X syndrome is examined. The first paper was prepared as if for submission to the journal *Behavior Modification*. The second paper was prepared as if for submission to the journal *American Journal on Intellectual and Developmental Disabilities*. Finally, the public domain briefing paper is an accessible summary of the main findings of the empirical paper.

Volume II contains five clinical practice reports relating to clinical work conducted whilst on placement in a learning disability, paediatric psychology and two adult mental health services. The first report provides a behaviour-analytic and a cognitive formulation of self-injurious behaviours displayed by an 18 year-old woman with severe intellectual and developmental disabilities, as well as of parental responses to her behaviour. Secondly, an evaluation of the role of Psychological Wellbeing Practioners within a primary care Improved Access to Psychological

Therapies service is presented. The third report is a single-case AB design, which provides an evaluation of a cognitive-behavioural treatment of a long-standing vomit phobia in an 18 year old woman. The fourth report is a case study of a 12 year old boy with eczema, in which a cycle of reflected shame served to precipitate and maintain high levels of distress for both the child and his mother. The final report is a case study of a 46 year old man with a long-standing, pervasive and debilitating experience of shame and self-attacking, which was assessed, formulated and treated using Compassion Focused Therapy. This was presented orally and as such, a one-page summary of this work is included.

DEDICATION

This thesis is dedicated to my beautiful family; Donna, Joseph and Ewan. You bring me so much happiness, love, and laughter…without which none of this work would have been possible.

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CHAPTER 1: LITERATURE REVIEW

THE MOTIVATING OPERATION AND NEGATIVELY REINFORCED PROBLEM BEHAVIOUR. A SYSTEMATIC REVIEW

Abstract

Motivating operations (MOs) exert value- and behaviour-altering effects on problem behaviour. To date, there has been no systematic review of the literature regarding the influence of MOs on negatively reinforced problem behaviour. The current review adopted a systematic strategy to identify and review papers relevant to this area published between 1999-2011. Fifty nine papers were identified that met inclusion criteria for the review. Papers were grouped according to themes and reviewed to: 1) identify recent trends in the literature, 2) provide a critique of the methodological strengths and weaknesses of the field, 3) examine implications for the assessment, understanding and treatment of negatively reinforced problem behaviour and 4) provide suggestions for future research. A model of negatively reinforced problem behaviour is presented that emphasises the importance of the interaction between person and environment. [1]

[1] The review has been prepared as if for submission to the journal *Behavior Modification*.

Introduction

Problem behaviours, such as aggression or self-injury, can exert a deleterious impact on quality of life. Such behaviours, which occur in 5-19% of people with intellectual and developmental disabilities (Borthwick-Duffy, 1994; Emerson et al., 2001; Joyce, Ditchfield, & Harris, 2001), are associated with a range of negative consequences including: social isolation (Robertson, Emerson, Gregory et al., 2001; Robertson et al., 2005), limited opportunities for choice or engagement in meaningful activity (Robertson, Emerson, Hatton et al., 2001; Mansell, 1995), and high levels of environmental restriction, sometimes resulting in physical or emotional abuse (Rusch, Hall, & Griffin, 1986).

Whilst a range of perspectives have been adopted to try to understand such behaviours, the operant model (see Oliver, 1995 for example) is the dominant paradigm for assessment, formulation and treatment. Within this model, problem behaviours are understood as behavioural adaptations to the antecedent and consequent conditions that arise within an individual's internal or external environment. Problem behaviours have been shown to be maintained by both positive and negative reinforcement processes (Hanley, Iwata, & McCord, 2003) and interventions based on this understanding have been demonstrated repeatedly to be effective in reducing such behaviours (Scotti, Evans, Meyer, & Walker, 1991).

Negative Reinforcement

Problem behaviours maintained by negative reinforcement have been the subject of considerable study over the past 30 years (e.g., Carr, Newsom, & Binkoff, 1976, 1980; Iwata, 1987). To quote Iwata (1987):

The process of negative reinforcement typically involves the removal, reduction, postponement, or prevention of stimulation; these operations strengthen the response on which they are contingent (p. 362).

With the advent of functional analytic methodologies has come increased conceptual and technological precision and an increased ability to isolate various aspects of the three-term contingency that serve to influence problem behaviour (Carr, 1994; Mace, 1994). Such advances have led not only to an improved understanding of the processes that influence negatively reinforced problem behaviours but also have demonstrated how such processes can be utilised to facilitate treatment (Hanley, Iwata, & McCord, 2003).

Motivating Operations

One part of the antecedent contingency to have received attention in recent years has been that of the motivating operation (MO; Laraway, Snycerski, Michael, & Poling, 2003; Michael, 1982, 1993, 2000). The MO refers to any event, or stimulus change that momentarily alters: a) the *value* of a particular stimulus as a source of reinforcement or punishment and b) the probability of *behaviours* that have been associated historically with such consequences[2]. For problem behaviours maintained by negative reinforcement, the onset of an MO establishes (or abolishes) the reinforcing value of escape from or avoidance of a given stimulus (such as

[2] MOs can be either *unconditioned* (i.e., result from the individual's phylogenic history), as in the deprivation of primary types of reinforcement (such as food, water or sexual activity) or *conditioned* (i.e., result from the individual's ontogenic history). It is beyond the scope of the current paper to provide a lengthy review of the concept of the MO and readers are referred to papers by Michael (1982; , 1993), and Langthorne and McGill (2009) for further description of the different classes of unconditioned and conditioned MOs and their influence on operant behaviour.

attention, pain or a demand) and evokes (or abates) behaviours associated with such consequences in the past. According to Iwata (1987), the defining feature of a negative reinforcement contingency is whether the change from an antecedent to consequent condition results in a reduction in aversive stimulation (p. 365). From this perspective, the extent to which a behaviour-consequence contingency constitutes negative reinforcement is dependent on the antecedent condition that precedes it.

A series of review papers (e.g., Hanley, Iwata, & McCord, 2003; Iwata, Smith, & Michael, 2000; McGill, 1999; Smith & Iwata, 1997; Wilder & Carr, 1998) have examined the influence of MOs on problem behaviour maintained by various sources of reinforcement. However, to date there has not been a systematic review of the literature on MOs focused exclusively on negatively reinforced problem behaviour. Two previous reviews of the influence of antecedent events on problem behaviours maintained by escape have been conducted (Carbone, Morgenstern, Zecchin-Tirri, & Kolberg, 2007; Miltenberger, 2006). However, both were limited to the analysis of negatively reinforced behaviours occurring in the context of instructional activities and neither reported a systematic methodology to either the identification or review of studies. Given the growing number of studies conducted within this field, a systematic approach towards the identification and review of papers is required to provide a more comprehensive account of the literature and facilitate a more rigorous assessment of its status.

The aims of the current review are to: a) identify trends over time in the study of MOs for negatively reinforced problem behaviour by comparing recent studies against those included in seminal reviews of the area (McGill, 1999), b) provide a summary of existing research for the assessment and treatment of MOs for negatively reinforced problem behaviour, c) identify the

strengths and limitations of existing methodologies used to assess MOs in negatively reinforced behaviour and d) provide recommendations for further research.

Methodology

Search Strategy

A systematic methodology using two separate search strategies was adopted to identify papers. In the initial strategy all empirical papers related to negatively reinforced problem behaviour were identified using all possible combinations of the following search terms: ('Avoidance' OR 'Escape' OR 'Negative Reinforcement') AND ('Behaviour Problems' OR 'Aggressive Behaviour' OR 'Self Destructive Behaviour' OR 'Behaviour Disorder') using the search engines *PsychInfo* and *Web of Science*.

As a number of key studies on MOs did not make explicit reference to negatively reinforced problem behaviour in the abstract or search terms, a second search strategy was used. In this search all studies that cited key MO conceptual papers (Laraway, Snycerski, Michael, & Poling, 2003; Michael, 1982, 1993, 2000) were identified. Studies that included an individual with problem behaviour maintained, at least in part, by negative reinforcement were then selected for further review.

The reference sections of all papers identified via each search strategy, as well as from previous reviews of the MO literature, were also searched to identify any related papers that may have fit the above criteria.

Inclusion and Exclusion Criteria

All papers identified in this initial search were reviewed by hand to determine whether the following inclusion criteria were met: 1) the study allowed for an inference that the problem

behaviour was, at least in part, negatively reinforced. This was based on indirect, descriptive or experimental functional analysis for all topographies of problem behaviour, with the exception of food refusal, in which studies were included that inferred behavioural function based on the nature of the topography of the behaviour[3], 2) the study included an assessment of the influence of an antecedent variable on the negatively reinforced problem behaviour and reported on the direct observation of the target behaviour. Studies of a correlational nature were only included if a within-subject experimental manipulation was not deemed possible due to the nature of the variable under study (i.e., presence of a specific biological condition). In cases where an experimental manipulation could reasonably have been expected to have been conducted and was not, then such studies were excluded from review. Papers were excluded if they did not include the assessment of an antecedent variable or were not focused on problem behaviour (e.g., focused on compliance only).

A body of studies that examined the effects of providing positive forms of reinforcement for compliance whilst maintaining an escape contingency for problem behaviour, best exemplified by Lalli et al (1999) was excluded from the review. Whilst a MO account of such findings is possible (see Fisher et al., 2005 for example), the primary interpretation of these findings has been made in terms of choice responding between two concurrent operants (Lalli et

[3] In relation to food refusal, only a handful of studies have included functional assessment data and the use of experimental functional analysis methodology has been a relatively recent development in this area. More typically a negative reinforcement function has been inferred via behavioural topography (by definition). Whilst there are a host of problems associated with relying on this inference, it was deemed important to include this body of studies in the current literature review, as prior reviews of negatively reinforced probalem behaviour have not included food refusal.

al., 1999). Studies that examined effects of this experimental manipulation, without providing further analysis to support an MO interpretation, were therefore excluded.

Historically, the function of negatively reinforced problem behaviour has been assessed by the use of a demand condition, whereby either the individual is presented with an task demand and the demand is removed contingent upon the occurrence of a target behaviour, as in an ABC analysis (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994), or the individual is presented with a non-preferred demand in the context of reduced attention and no programmed consequences are provided for the behaviour, as in an AB analysis (Carr & Durand, 1985). As the influence of this specific manipulation has been well documented across hundreds of studies (see Hanley, Iwata, & McCord, 2003 for a review), papers that included solely a demonstration of either of the above methodologies, without the use of any additional variants, were excluded.

Multi-component interventions that involved the manipulation of multiple variables at the same time were also excluded, in cases where the independent effects of an antecedent variable could not be inferred.

The review included papers published between 1999 and 2011. Studies pre-dating 1999 have previously been comprehensively reviewed in seminal papers on MOs (McGill, 1999; Smith & Iwata, 1997). The current review therefore included all studies that met inclusion criteria that were published between 1999 and 2011.

Methodology for Reviewing the Quality of the Evidence Base

In accordance with the wider move towards evidence-based practice (Kaiser & McIntyre, 2010), the current review aimed to evaluate studies against recognized criteria for the evaluation

of single-case experimental design methodology (Kratochwill et al., 2010). These criteria are described in Table 1.

Table 1. *Summary of Criteria for Single-Case Designs that Meet Evidence Standards (With or Without Reservation). What Works Clearing House (Kratochwill et al., 2010).*

Standard.	Outcome.
1. The IV (intervention) must be systematically manipulated[4] with the researcher determining when and how the IV conditions change	Meets evidence standards *with* or *without* reservations (indicate which)[5]/Does not meet evidence standards
2. Each outcome variable must be measured systematically over time by more than one assessor and the study needs to collect inter-assessor agreement in each phase and on at least twenty percent of the data points in each condition (e.g., baseline, intervention) and the inter-assessor agreement must meet minimal thresholds.	Meets evidence standards *with* or *without* reservations (indicate which)[6]/Does not meet evidence standards
3. The study must include at least three attempts to demonstrate an intervention effect at three different points in time or	Meets evidence standards *with* or *without* reservations (indicate which)[7]/Does not meet evidence standards

[4] Added by author "in cases where manipulations are made that could introduce a confounding variable (e.g., use of non-trained individuals as interventionists) then efforts to control for these potential confounds should be made (e.g., use of treatment integrity data). In cases where standard not met then reject"

[5] If standard not met then reject

[6] Must have collected for at least 20% of intervals, for each case on each outcome variable. If not then reject. Minimum acceptable range from 0.8-0.9 for percentage agreement and 0.6 if using kappa statistic.

[7] If standard not met then reject. Examples of designs not meeting this standard include AB, ABA, and BAB designs.

	with three different phase repetitions.	
4.	For a phase to qualify as an attempt to demonstrate an effect, the phase must have a minimum of three data points.	Meets evidence standards *with* or *without* reservations (indicate which)[8]/Does not meet evidence standards

Results

A total of 4,440 papers were identified through the initial two search strategies and an additional 20 papers were identified by searching the reference lists of other papers. Following a review of the title and abstracts of all papers, a total of 92 papers were selected for further detailed review. Of these 92 studies, 59 met selection criteria and formed the basis of the review. From these 59 papers, a total of 70 different experimental manipulations were abstracted and compiled into Tables 2-9.

Eight studies were identified that examined the role of biological variables (e.g., genetic syndromes, health conditions, medication) in negatively reinforced problem behaviour (see Table 2). Seventeen studies were identified which examined antecedent conditions, (other than the onset of academic demands) that appeared to occasion negatively reinforced problem behaviour

[8] To *Meet Standards* a reversal /withdrawal (e.g., ABAB) design must have a minimum of four phases per case with at least 5 data points per phase. To *Meet Standards with Reservations* a reversal /withdrawal (e.g., ABAB) design must have a minimum of four phases per case with at least 3 data points per phase. Any phases based on fewer than three data *points cannot* b*e used to demonstrat*e existence or lack of an effect. To *Meet Standards* a multiple baseline design must have a minimum of six phases with at least 5 data points per phase. To *Meet Standards with Reservations* a multiple baseline design must have a minimum of six phases with at least 3 data points per phase. Any phases based on fewer than three data points *cannot* b*e used to demonstrat*e existence or lack of an effect. An alternating treatment design needs *five repetitions* of the alternating sequence to *Meet Standards.* A design with four repetitions would *Meet Standards with Reservations*, and a design with fewer than four repetitions *Does Not Meet Standards.*

and their parameters (see Table 3). Four studies were examined that influenced the effect of making pre-session manipulations prior to the onset of instructional demands (see Table 4); 11 studies were identified that examined the effect of altering the mode of demand presentation (see Table 5); eight studies were identified that investigated the influence of manipulating the difficulty of instructional demands (see Table 6); four studies were identified that examined the influence of manipulating choice or predictability of instructional demands (see Table 7); five studies were identified that investigated the influence of altering the schedule of instructional demands (see Table 8) and thirteen studies were identified that examined the influence of adding potential sources of positive reinforcement to an aversive context (see Table 9).

Of these 70 experimental manipulations, 39 were deemed to meet all four experimental standards (with or without reservations) proposed by Krachtochwill et al (2010) for one or more participants; fifteen were deemed to meet at least three of the experimental standards, nine were deemed to meet at least two of the experimental standards, two studies were deemed to meet at least one of the experimental standards, and one study was included which failed to meet any of the experimental standards. Four studies were deemed exempt from this methodological review process as they were descriptive in nature, and included the evaluation of variables that could not reasonably be examined experimentally within a single-case experimental design paradigm.

Biological Influences

The concept of the MO has proved important in helping to bridge the historical divide between the biological and operant sciences (Langthorne, McGill, & O'Reilly, 2007; Oliver, 1993). Biological variables appear to play a critically important role in influencing the development and subsequent maintenance of escape-maintained problem behaviour. In his review, McGill (1999) identified a handful of studies to have examined the role of sleep, allergies and physical illness as MOs for escape-maintained problem behaviours (Horner, Day, & Day, 1997; Kennedy & Meyer, 1996; O'Reilly, 1995). Whilst conceptual arguments had been made for the influence of more enduring genetic influences on the reinforcing value of specific sources of reinforcement (McGill, 1999; Oliver, 1993), there was an absence of empirical evidence to support these hypotheses.

Over the past decade there have been some considerable developments in this field (see Table 2). First, in relation to the influence of genetic disorders there has been considerable conceptual (Langthorne & McGill, 2008) and empirical (Tunnicliffe & Oliver, 2011) appreciation of their contribution to the phylogeny and ontogeny of problem behaviour. In relation to negatively reinforced problem behaviour, two studies were identified that utilized aggregate single-case design methodology to provide preliminary evidence to support possible elevated rates of negatively reinforced problem behaviour in both fragile X syndrome (Langthorne et al., 2011) and Angelman syndrome (Strachan et al., 2009). Whilst some evidence, using indirect methods of functional assessment, exists to support within- and between-subject differences in the distribution of specific behavioural functions (Langthorne & McGill, 2012), there is a need

for large-scale, group comparison studies that employ experimental functional analytic methodologies.

Another important avenue in this line of research lies in the analysis of specific biological, cognitive or behavioural characteristics associated with particular syndromes and their interaction with the environmental conditions that give rise to problem behaviour. In the current review, one study was identified that demonstrated a relationship between hyperacusis and problem behaviour occurring under demand conditions for a child with Williams syndrome (O'Reilly, Lacey, & Lancioni, 2000). Specifically, whilst pain-related behaviours occurred when a loud noise was present, problem behaviour was only evoked when demands were combined with loud noise. The use of ear plugs in this specific context reduced escape-maintained problem behaviours and pain-related behaviour. Single-case design methodology is particularly well suited towards meeting the needs of this type of research and further research is important if interventions are to be identified that help to meet the needs of people with genetic conditions associated with intellectual disabilities and problem behaviour.

There has been continued investigation of the role of specific physiological variables on negatively reinforced problem behaviour, including sleep deprivation (O'Reilly & Lancioni, 2000) and menses (Carr, Smith, Giacin, Whelan, & Pancari, 2003). The study by O'Reilly and Lancioni is particularly noteworthy for demonstrating an interaction between sleep deprivation and an increase in a specific member of a response class hierarchy (self-injurious behaviour). The interaction between MOs and the 'price' individual's will pay (i.e., response cost) to access specific sources of reinforcement (such as escape) has received scant attention and may be an important parameter that influences the *matching law* (Herrnstein, 1961) and the distribution of

responses when concurrent operants are available. This should be a priority for future basic and applied research on negatively reinforced problem behaviour.

Interestingly, there appears to be a preponderance of studies that have demonstrated a relationship between fluctuations in health conditions and negatively reinforced problem behaviour, as opposed to behaviours that serve other behavioural functions (Kennedy & Becker, 2006). It is unclear whether such a specific relationship exists and the nature of the mechanisms that could underpin such a relationship. Further examination of these questions could offer important advances in understanding of negatively reinforced problem behaviour and the pathways that underpin it.

Finally, examination of the influence of certain medications has been shown to influence problem behaviour maintained by negative reinforcement. Two studies were identified that investigated the influence of risperidone on behavioural function during a cross over medication trial (Crosland et al., 2003; Zarcone et al., 2004). These studies suggested relatively idiosyncratic effects on escape-maintained problem behaviour across different individuals. Both were, however, hampered by difficulties with experimental control. A well controlled study, reported by Kelley, Fisher, Lomas and Sanders (2006), noted a shift in response allocation from problem behaviour to compliance following the introduction of amphetamine for a child with ADHD. This appears to provide an interesting paradigm through which the influence of specific medications can be investigated.

These findings have important implications for understanding the development of problem behaviour and the ontogeny of specific behavioural functions. Equally, they have clear

implications for the assessment of escape-maintained problem behaviour. In cases where there is variability in problem behaviour then the influence of fluctuations in medications and health conditions should be considered as a potential contributory factor. In cases where their role is implicated then the treatment and amelioration of any discomfort should be prioritized (Carr & Blakeley-Smith, 2006). The findings of O'Reilly, Lacey and Lancioni (2000) highlight the importance of developing an understanding of phenotype-environment interactions in order to develop environments that are matched to the needs of individuals with specific phenotypes. Despite the advances that have been made in this area in recent years, there remains scant research that has investigated the mechanisms by which various biological variables serve to increase (or reduce) the probability of problem behaviour. As such, greater attention is required to delineate precisely the pathways between the onset of a particular biological variable and variability in problem behaviour.

Table 2. *Studies that Demonstrate the Influence of Biological Variables on Negatively Reinforced Problem Behaviour.*

Study	Participants (age, diagnoses)	DV	Functional assessment[9]	IV	Design	Experimental standards met	Findings
Kelley, Fisher, Lomas, & Sanders, (2006)	*Jake* (11yrs, moderate ID, ADHD)	Destructive behaviour ***	ABC	1)Placebo Vs. 2)Amphetamine during demand condition	Reversal ABABABABA (double blind design)	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Destructive behaviour reduced and higher levels of compliance under amphetamine
Crossland et al (2003)	*Reggie* (6 yrs, autism, profound ID, and fragile X syndrome) **	Destructive behaviour	ABC	Functional analysis conducted during trial of risperidone	Reversal ABA design with embedded multi-element design	Standard 1-N Standard 2-Y Standard 3-N Standard 4-N	Risperidone correlated with reduction in escape maintained problem behaviour and increase in compliance, no influence on

*Data from study reported elsewhere in review, ** term 'intellectual disability' used instead of term used in paper (e.g., 'mental retardation'), ***data on compliance also reported in the study.

[9] ABC analysis refers to an experimental comparison between at least one test and control condition whereby both antecedents and consequence conditions are manipulated. Antecedent analysis refers to an experimental comparison between at least one test and control condition whereby antecedents only have been manipulated. Descriptive assessment refers to an observational, non-experimental assessment of problem behaviour. Indirect assessment refers to assessments informed by caregivers/teachers. Function inferred by topography refers to studies whereby behavioural topography has been used to make an inference regarding behavioural function (i.e., food refusal). In studies where multiple methods of functional assessment were included then the method with the highest level of control is reported herein.

							tangible
O'Reilly, Lacey, & Lancioni, (2000)	*Eilis* (5yrs, Williams syndrome, hypercalcemia , moderate ID)	Problem behaviour (and pain behaviour)	ABC	Functional analysis introduced under 1) No noise, Vs. 2) Noise, Vs. 3) Noise with ear plugs	ABABCBC reversal design with embedded multi-element	Standard 1-Y Standard 2- Y Standard 3-Y Standard 4-Y	Problem behaviour at higher rates under high noise. Pain behaviours were present when noise high and no plugs
O'Reilly & Lancioni (2000)	*Sarah* (4yrs, moderate ID)	SIB, aggression	ABC	1)Demand conditions after nap Vs. 2) Demand conditions no nap	Naturally occurring reversal ABAB	Standard 1- N Standard 2-Y Standard 3- Y Standard 4- N	Sleep deprivation increased the occurrence of SIB, whilst having no apparent effect on aggression. Both members of same response class
Carr, McLaughlin, Giacobbe-Grieco, & Smith (2003)	Four females with moderate-severe ID (age range 26 yrs-31yrs)**	Problem behaviour	Antecedent	1) Menses plus demands Vs.2) Menses without demands Vs.3) No menses plus demands Vs. 4) No menses, no demands.	Multi-element embedded within a naturally occurring reversal	Standard 1-N Standard 2-Y Standard 3-Y Standard 4-N	All participants higher levels of problem behaviour occurring in the menses plus demands condition of the analysis

Aggregate Studies

Langthorne et al (2011)	8 boys with fragile X syndrome (age range; 8-15 yrs)	Problem behaviour	ABC	Examined function served by problem behavioural of group of 8 children with FXS	Descriptive study	N/A	5/8 children had one form of escape-maintained behaviour
Strachan et al (2009)	7 boys and 5 girls with Angelman syndrome (age range 5-11yrs)	Aggression	Antecedent	Examination of function of problem behaviour of 12 children with Angelman syndrome	Descriptive study	N/A	8 of the 10 participants who displayed aggressive behaviour did so in the high attention condition and four out of ten in demand condition.
Zarcone et al (2004)	8 children and 5 adults with autism and other DD	Destructive responses	ABC	Functional analysis conducted during a medication trial of risperidone	Multi-element design within a double-blind, placebo-controlled trial	N/A	Idiosyncratic responses for escape-maintained behaviours across different individuals

Negative Sources of Negative Reinforcement

From the advent of functional analytic methodology (Carr, Newsom, & Binkoff, 1976, 1980; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994), the presentation of instructional academic demands has been used as the standard MO to test for the presence of a negative reinforcement contingency. In his review of motivating operations, McGill (1999) identified only a handful of studies that tested for alternative sources of motivation for negatively reinforced problem behaviour, specifically for the onset of social attention (Taylor & E. G. Carr, 1992; Taylor & Carr, 1992) and ambient noise (O'Reilly, 1997).

The current review identified strong evidence for the role of social contact as a source of aversive stimulation. Both Hagopian, Wilson and Wilder (2001) and Tiger et al. (2009) reported on the use of an 'escape from attention' condition as a variant to the standard functional analysis. In both studies the MO in this condition comprised the continuous presentation of social attention followed by 30s of escape contingent on the problem behaviour and was implemented after elevated rates of problem behaviour had been found in the 'Play' condition of a prior functional analysis. As demonstrated by Oliver, Oxener, Hearn and Hall (2001), however, for some individuals it may not be social attention per se but rather social proximity that proves to be the critical source of aversive stimulation associated with social contact.

Such factors may conceivably influence problem behaviour occurring in the context of instructional demands. Moore and Edwards (2003) identified two participants who showed higher levels of escape-maintained problem behaviour in conditions associated with high levels of non-contingent attention during school work. A subsequent analysis revealed that providing praise for engagement was associated with higher rates of problem behaviour and lower rates of

engagement in comparison to attending to disengagement. This suggests that for individuals who find attention aversive, it may be the attention-component of demand presentation, rather than the demand itself, that evokes escape-maintained problem behaviour.

Other studies have provided further evidence to show that problem behaviours occurring in the context of instructional demands may not necessarily indicate that the instructional sequence is the aversive component of the demand. McCord, Thomsen and Iwata (2001) completed a functional analysis to identify the aversive aspects of transitions between activities. The authors found that requests to change location, irrespective of the nature of the ongoing or subsequent task, motivated problem behaviour for both participants. Likewise Hagopian et al. (2007) demonstrated that requests to complete an instructional demand may be aversive because of the interruption to ongoing activities. It may be that in such cases, the ongoing activity functions as a form of transitive CMO, in that its onset establishes another stimulus (i.e., the demand) as aversive and evokes behaviours associated with the termination of the demand. In the absence of the ongoing activity one would expect that the demand would not hold its aversive properties.

Further evidence was also found for the presentation of food as an MO for negative reinforced problem behaviour (Bachmeyer et al., 2009; LaRue et al., 2011; Piazza et al., 2003). A handful of studies have provided a more fine-grained analysis of such sources of negative reinforcement. Rivas, Piazza, Patel, and Bachmeyer (2010) for example demonstrated the distance between a spoonful of food and the mouth acted as a form of MO. This assessment was then used to direct an intervention directed at gradually fading the distance between the spoon and lips of a participant, under escape extinction conditions. Similarly, other manipulations, such

as preference for specific types of food (Levin & Carr, 2001) have been shown to alter the value of escape in problem behaviours associated with food refusal.

Whilst the presence of noise has previously been demonstrated to form a general class of MO, more recent studies have provided a more fine-grained analysis of the specific type of sounds that can establish specific noises as aversive (Buckley & Newchok, 2006; McCord, Iwata, Galensky, Ellingson, & Thomson, 2001).

These findings (shown in Table 3) have a number of implications for the assessment and treatment of problem behaviour. First, it is important to assess a broader range of stimuli other than instructional demands when testing for a negative reinforcement contingency. Failure to do so may result in a Type II error (i.e., the presence of a negative reinforcement contingency may be missed when it is present). Pre-assessment, in the form of indirect or direct observation, has been shown to be critical in helping to identify a range of potentially aversive stimuli to include in an assessment (e.g., Roscoe, Rooker, Pence, & Longworth, 2009). Indeed, the current review identified a number of studies that incorporated a broader assessment of demands to assess for possible sources of aversive stimulation (Baker, Hanley, & Mathews, 2006; Long, Hagopian, DeLeon, Marhefka, & Resau, 2005; Roscoe, Rooker, Pence, & Longworth, 2009).

Equally, it is important to be responsive to the specific results of a functional analysis. Problem behaviours occurring in the control condition of a functional analysis should prompt the use of a condition to test the role of social contact as an aversive stimulus. Also, it should not be assumed that simply because problem behaviour occurs in the general context of demands that it is the instructional demand that necessarily functions as the relevant MO. Within-session

analyses may be required to help identify the aversive aspect of the demand context (Roane, Lerman, Kelley, & Van Camp, 1999). This would be especially indicated in situations where an individual continues to display problem behaviour irrespective of within-session fluctuations in the presence of demands. For example, within-session analyses of MOs could be used to determine whether the conditional probability of problem behaviour increases following the delivery of praise for compliance, which may indicate that it is the social contact aspect of demands that is aversive. In cases where such a relationship was found then modifications to the prompting procedures could be made that encouraged engagement with the instructional sequence, whilst minimizing aversive social contact. Greater use of indices of negative affect may help to indicate specific aspects of an environmental context that an individual finds aversive and, clinically, would help to guide efforts at early intervention, potentially before the onset of problem behaviour (Lindauer, DeLeon & Fisher, 1999). Finally, there are comparatively few studies to have provided fine-grained MO analyses of these forms of aversive stimulation; this would seem to be a priority for future research due to the implications such analyses have for the treatment of problem behaviours maintained by alternative sources of negative reinforcement.

Table 3. *Studies that Demonstrate the Influence of MOs for Novel Sources of Negative Reinforcement.*

Study	Participants (age, diagnoses)	DV	Functional assessment [10]	IV	Design	Experimental standards met	Findings
LaRue et al (2011)	*Lauren* (2yrs) *George* (5yrs) *Carl* (18mths) *Charles* (5yrs) *Frank* (21mths)	Inappropriate behaviour ***	ABC	1)Control Vs. attention Vs. 2)Escape conditions during mealtime functional analysis	Pairwise multi-element design (except Charles, ABAB reversal)	Standard 1- Y Standard 2- Y Standard 3- Y Standard 4- Y (Frank, Charles), R (Carl), N Lauren, George	All presented behaviour maintained by escape from food
Rivas, Piazza, Patel & Bachmeyer(2010)	*David* (4yrs, failure to thrive) *Ashley* (5 mths, failure to thrive) *Oliver* (10mths,	Inappropriate mealtime behaviour ***	ABC	Phase 1:1)Spoon at distance Vs.2) Spoon at lips Phase 2: 1)Fading distance Vs. 2) Fading + Extinction. Vs. 3) Extinction Vs.	Phase 1: Alternating treatment Phase 2: Reversal + alternating treatments	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Rates higher in condition where spoon placed to lips.Fading plus escape extinction more effective than alternatives.

[10] ABC analysis refers to an experimental comparison between at least one test and control condition whereby both antecedents and consequence conditions are manipulated. Antecedent analysis refers to an experimental comparison between at least one test and control condition whereby antecedents only have been manipulated. Descriptive assessment refers to an observational, non-experimental assessment of problem behaviour. Indirect assessment refers to assessments informed by caregivers/teachers. Function inferred by topography refers to studies whereby behavioural topography has been used to make an inference regarding behavioural function (i.e., food refusal). In studies where multiple methods of functional assessment were included then the method with the highest level of control is reported herein.

	failure to thrive)			Ext + fading (Oliver)	(Oliver)		
Tiger, Fisher, Toussaint, & Kodak (2009)*	*Jimmy* (13yrs, autism)	Aggression	ABC	1)Social escape Vs. 2) No interaction	Multi-element	Standard 1- Y Standard 2- Y Standard 3- Y Standard 4- R	Demonstrated an escape from attention function
Roscoe, Rooker, Pence, & Longworth (2009)	*Steve* (14yrs, autism) *Jill* (10yrs, Smith-Magenis syndrome) *Candace* (10yrs, autism) *Tyler* (22yrs, profound intellectual disabilities)**	Aggression (Steve, Candace) Property destruction (Tyler) SIB (Jill) ***	ABC	Phase 1: Demand assessment Phase 2: 1)Attention Vs. 2)Control Vs.3) Low p demand Vs. 4)High p demand conditions	Multi-element design	Standard 1- Y Standard 2- Y Standard 3- Y Standard 4- R [Steve and Jill], N [Candace and Tyler]	Low p demands evoked higher rates for 3 participants.
Bachmeyer et al (2009)	*Mathew* (4yrs, DD) *Ella* (4yrs, DD) *Tyler* (5yrs, typically developing) *Savannah* (3yrs, DD)	Inappropriate mealtime behaviour ***	ABC	1)Control Vs. 2)Attention Vs. 3)Escape conditions during food presentation.	Pair-wise, multi-element design.	Standard 1- Y Standard 2- Y Standard 3-Y Standard 4- Y [Tyler & Savannah], N [Mathew & Ella]	Behaviour at higher rates in the escape and attention conditions

Hagopian Bruzek, Bowman, & Jennett (2007)	*Perry* (12yrs, moderate ID, autism) *Maxwell* (6yrs, moderate ID, autism), *Kelly* (12yrs, autism,) **	Problem behaviour	ABC	Perry and Maxwell: 1)Control condition Vs.2) 'Do' mands (interrupted ongoing activity) with escape contingency Kelly. 1)'Do' requests Vs. 2) 'Don't requests (e.g., 'you can't sit here')	Multi-element (Perry, Kelly), reversal (Maxwell)	Standard 1-Y Standard 2–Y Standard 3-Y Standard 4-Y	Problem behaviour at higher rates in the interruption condition. 'Do' and 'don't' requests were found to evoke problem behaviour.
Baker, Hanley & Mathews (2006)*	*Participant* (96 yrs, Alzheimer's dementia)	Aggression	ABC	Phase 1: 1) Bathroom routine Vs. 2)Recreational routine. Phase 2. 1)Escape Vs. 2)Attention Vs. 3)Control during bathroom routine	Multi-element	Standard 1- Y Standard 2- Y Standard 3- Y Standard 4- Y [phase1], R [phase 2]	Phase 1: Aggression occurred only in bathroom routine. Phase 2: Elevated rates in escape and attention condition.
Buckley & Newchok (2006)	*Billy* (7yrs, pervasive DD)	Disruption	ABC	Leisure Vs. Music from a tape with escape Vs. Music from a CD with escape	Multi-element	Standard 1- Y Standard 2- Y Standard 3-Y Standard 4- Y	Higher levels of disruption during tape condition, irrespective of music genre.
Piazza et al (2003)	5 females (aged 1-2yrs) 10 males (aged	Inappropriate mealtime behaviour	ABC	1)Control Vs. 2)Tangible Vs. 3)Escape Vs.	Reversal design	Standard 1- Y Standard 2- Y Standard 3- N	9 of 10 children who displayed problem behaviour found to be at

	1-7yrs)			4)Attention conditions		Standard 4- Y	least in part be escape-maintained
Moore & Edwards (2003)*	*Edgar*, (9yrs, severely emotionally disturbed) *Morris*, (7yrs, cognitive and academic abilities in average range) *Jacob,* (17yrs) *Robert* (7 yrs)	Problem behaviour ***	ABC	Phase 2.1) Low attention/easy demand Vs. 2)Low attention/difficult demand Vs. 3)High attention/easy demand Vs. 4)High attention/difficult demand. Phase 3. 1)Praise for engagement Vs. 2)Encouragement for disengagement Vs. 3)Reprimand for disengagement.	Multi-element	Standard 1-Y Standard 2- Y Standard 3- Y Standard 4- R [phase 3, Edgar], N [all other data sets]	Phase 2. Edgar and Morris higher problem behaviour in high attention conditions. Phase 3. Edgar and Morris, higher problem behaviour when praise provided for engagement. Converse when the teacher attended to disengagement.
Hagopian, Wilson & Wilder (2001)	*Preston* (6-yrs, autism and mild ID)**	SIB, aggression, disruption, spitting	ABC	Phase 1. Functional analysis. Phase 2. Functional analysis with 'escape from attention condition'.	Multi-element	Standard 1- Y Standard 2- Y Standard 3- Y Standard 4- Y	Phase 1. High rates of problem behaviour in play condition. Phase 2. Showed elevated rates of problem behaviour in escape from attention

							condition.
McCord, Iwata, Galensky, Ellingson, & Thomson (2001)	*Debbie* (43yrs, autism, profound ID) *Sarah* (41yrs, severe ID) **	Problem behaviour	ABC	Phase 1: 1)Noises identified as potential EOs Vs. 2)Control (white noise, & man talking in conversational tones). Escape contingency. Phase 2: Debbie and Sarah. 1)Noise condition Vs. 2)Play Vs. 3) No interaction conditions. Phase 3: 1) Extinction alone Vs.2) Stimulus fading + extinction	Multi-element (Phase 1 and Phase 2) Multiple baseline across participants (Phase 3)	Standard 1- Y Standard 2- Y Standard 3- N (phase 3) Standard 4- N (phases 1, 3)Y (phase 2, Sarah); R (Debbie)	Phase 1: Specific noises evoked problem behaviour for Debbie and Sarah. Phase 2: Verified that problem behaviour maintained by escape from noise. Phase 3: Stimulus fading + extinction reduced the problem behaviour for Debbie. DRO needed for Sarah.
McCord, Thomson & Iwata (2001)	*Hayden* (27yrs, profound ID) *Michael* (38yrs, profound ID) **	SIB	ABC	1)Activity initiation, no location change Vs. 2)Activity initiation, location change Vs. 3)Activity termination, no location change Vs. 4)Activity termination, location	Multi-element	Standard 1- Y Standard 2- Y Standard 3- Y Standard 4- Y	Hayden: SIB maintained by avoidance of location change Michael: as above, but also served the function of escape from ongoing tasks and

				change, Vs. 5)Location change, no activity			avoidance of task initiations
Moore, Edwards, Wilczynski & Olmi (2001)	*Participant 1* (4 yrs, boy) *Participant 2* (5 yrs, boy) *Participant 3* (9yrs, girl) *Participant 4* (9yrs, boy)	Problem behaviour ***	Antecedent	Care-giver presented 1)Free play Vs.2) High difficulty demand Vs. 3)Low difficulty demand Vs. 4)High attention Vs. 5)Low attention	Brief multi-element with ABAB reversal probes	Standard 1-Y Standard 2- Y Standard 3- Y Standard 4- N	Participants 3and 4 demonstrated higher levels of problem behaviour in high attention condition
Oliver, Oxener, Hearn, & Hall, (2001)	*Alex* (14 yrs, ID) **	Problem behaviours	Antecedent	1) Close proximity Vs. 2) 2m proximity	Alternating treatment design	Standard 1- Y Standard 2-Y Standard 3-Y Standard 4-Y	Differentiation between the conditions irrespective of social contact. Higher in close proximity.
Levin & Carr (2001)	*Jack* (6 yrs, ASD, IQ 37) *Luis* (5 yrs, ASD, IQ 56) *Manny* (7 yrs, ASD, IQ 40) *Bess* (6 yrs, ASD, IQ 47)	Problem behaviour ***	ABC	1)Preferred food Vs. 2) Non-preferred food. Escape contingency.	ABAB	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	High levels of problem behaviour and low levels of food consumption in non-preferred conditions
O'Reilly, Lacey, & Lancioni,	*Eilis* (5yrs, Williams syndrome,	Problem behaviour (and pain	ABC	No noise Vs. Noise Vs. Noise with ear plugs	ABABCBC reversal design with embedded	Standard 1-Y Standard 2- Y Standard 3-Y	Problem behaviour at higher rates under

(2000)	moderate ID)	behaviour)	multi-element	Standard 4-Y	demand = + high noise. Pain behaviours present when noise high and no plugs

Manipulation of Pre-Session Variables

There has been a relatively well established line of research, since the publication of McGill (1999), that has examined the influence of manipulating pre-session variables in order to examine the subsequent motivative effects on different sources of positive reinforcement (Edrisinha & O'Reilly, 2006; O'Reilly, 1999; O'Reilly et al., 2006; Roantree & Kennedy, 2006). However, the influence of such manipulations on behaviours maintained by escape from aversive stimuli has received comparatively little attention.

Only four studies identified examined such variables for escape maintained problem behaviours (see Table 4). Whilst pre-session contexts characterized by high levels of demands have been shown to act as an EO for subsequent escape-maintained problem behaviours occurring within a functional analysis (O'Reilly, Lancioni, & Emerson, 1999), the majority of studies have focused on examining the potential influence of alternative pre-session variables, some of which can be controlled experimentally by, for example, manipulating pre-session access to attention (McComas, Thompson, & Johnson, 2003) or preferred tangible items (Rispoli et al., 2011). Other studies have attempted to identify correlations between more temporally distal events (Ray & Watson, 2001) and negative reinforced problem behaviour. However, due to their correlational nature it is not possible to determine the extent to which such events serve to act as MOs. Given their likely influence such variables should be identified increasingly through indirect and direct methods of assessment and experimental analyses.

Table 4. *Studies that Demonstrate the Influence of Manipulating Pre-session Conditions as an MO for Negatively Reinforced Problem Behaviour.*

Study	**Participants (age, diagnoses)**	**DV**	**Functional assessment**	**IV**	**Design**	**Experimental standards met**	**Findings**
Rispoli et al (2011)	*Rusty* (9 yrs, autism)	Problem behaviour***	ABC[11]	1) Pre session access to tangibles Vs. 2) Pre-session no access during classroom instruction.	Multi-element	Standard 1-Y Standard 2-Y Standard 3- Y Standard 4- Y	More problem behaviour (and lower engagement) following pre-session no access conditions.
McComas, Thompson, & Johnson (2003)	*Stan* (10 yrs, mild to moderate ID) *Abe* (11yrs, autism and moderate to severe ID) *Ari* (12 yrs, Down Syndrome, and moderate ID)	Problem behaviour	ABC	1)Pre session attention Vs. 2) Pre-session no attention	Multi-element	Standard 1-Y Standard 2-Y Standard 3- Y Standard 4- Y (Abe), N (Stan, Ari)	No relation between pre-session manipulation and escape maintained behaviour

[11] Indicated behavior maintained by both access to tangibles and escape from instructional demands.

Ray & Watson (2001)	*Kevin* (5yrs, IQ in low-average functioning) *Arthur* (5yrs, average IQ functioning)	Aggression	ABC	Descriptive assessment of temporally distant events (TDE) on results of functional analysis. TDE for Kevin was waking late, for Arthur was nocturnal enuresis.	N/A	Descriptive study	Higher problem behaviour in escape condition on days in which the TDE was present
O'Reilly (1999)	*Jeff* (31yrs, severe ID)	SIB	ABC	1) Pre-session alone for 30 mins Vs. 2) Pre-session 30mins of demands	ABAB with embedded multi-element design	Standard 1-Y Standard 2-Y Standard 3- Y Standard 4- Y	Rates of escape-maintained behaviour higher following pre-session demands than pre-session alone conditions.

Altering the Mode of Demand Presentation

Studies have begun to discriminate between a demand (i.e., task that needs completing) and the prompting procedure that is used to support it (i.e., mode with which demands are presented). A number of studies (see Table 5) were identified that manipulated the specific prompting procedures used to support an individual in completing instructional demands. This appears to have been a relatively recent development and did not feature in the studies reviewed by McGill (1999). Three studies investigated the influence of providing different numbers of steps to a prompting procedure, with relatively idiosyncratic effects reported (Boelter et al., 2007; Crockett & Hagopian, 2006; Stichter, Sasso, & Jolivette, 2004; Tiger, Fisher, Toussaint, & Kodak, 2009). For example, Tiger et al. reported that problem behaviour reduced following the introduction of a graduated, 3-step prompting procedure, in comparison to a 1-step, verbal only prompting procedure, whereas both Boelter et al (2007) and Crockett and Hagopian (2006) report a contrasting pattern of results following similar manipulations in their studies. It would be of interest to see whether other factors, such as level of task difficulty, are related to such variability.

Other studies have demonstrated that manipulating the style with which prompts are delivered (Borrero, Vollmer, & Borrero, 2004a; Peyton, Lindauer, & Richman, 2005) and the way in which corrective feedback is provided (Ebanks & Fisher, 2003) can exert abolishing and abative effects on escape-maintained behaviour. For example, Peyton et al demonstrated that altering the directiveness of prompts (e.g., from "show me the *X*" to "I wonder where the *X* is") successfully reduced the occurrence of escape maintained problem behaviour in a 10 year old girl with autism. Similarly the verbal description used to describe an escape contingency has been

shown to alter the probability of escape-maintained problem behaviour (Northup, Kodak, Lee, & Coyne, 2004).

These studies are important in that they demonstrate that the task demand per se may not be the aversive feature of an instructional sequence but rather the prompting procedure used to support it may be. In applied settings, when an individual shows variation between different people presenting similar demands then it may be that differences in prompting procedures underpin such variability, as opposed to any feature of the task itself. Such variations may be important in accounting for the effects of factors such as 'rapport' on problem behaviour (Magito-McLaughlin & Carr, 2005). It seems important that this is explored further in applied contexts as successful compliance with instructional demands could be elicited by a simple change in prompting procedure rather than necessitating a change to the task itself. Research is required that helps to elucidate when these relations are operative in order to help ensure that this important distinction is not overlooked.

Table 5. *Studies that Examined the Influence of Altering the Mode of Demand Presentation as an MO for Negatively Reinforced Problem Behaviour.*

Study	**Participants (age, diagnoses)**	**DV**	**Functional assessment**	**IV**	**Design**	**Experimental standards met**	**Findings**
Tiger, Fisher, Toussaint, & Kodak (2009)*	*Carl* (10yrs, ID) **	Problem behaviours	ABC	1)3-step prompting procedure Vs. 2) Verbal prompting procedure.	ABAB reversal with embedded multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Problem behaviours at zero rates when 3- step prompting procedure adopted.
Butler & Luiselli (2007)*	*Michelle* (13yrs, autism)	Problem behaviour	ABC	Comparison of 5 different individuals presenting request associated with problem behaviour.	Multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-N	Problem behaviours more with individuals associated with longer learning history
Boelter et al (2007) * Phase 3	*James* (4 yrs, ADHD) *Marcus* (3 yrs, , ADHD, mild DD) *Beto* (6yrs, autism)	Disruptive behaviour ***	Indirect	1)1 step directives Vs.2) 3 step directives during preferred (James, Marcus) or non-preferred (Beto) tasks	Multi-element probe design with reversals	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-N	Beto. More disruptive behaviour and reduced accuracy under 3 step conditions. Marcus and James. No effect on disruptive behaviour,

							improved accuracy in 1 step conditions
Crockett & Hagopian (2006)	*Chuck* (19 yrs, Nager's Syndrome, mild ID) **	Destructive behaviour ***	ABC	1) 3-step prompting procedure Vs. 2) Verbal prompt at onset of the session	Multiple baseline (across tasks)	Standard 1-Y Standard 2-Y Standard 3- N Standard 4- Y	Reduction in destructive behaviour to zero in verbal prompt condition. Increase in number of tasks completed.
Peyton, Lindauer, & Richman, (2005)	*Suzie* (10yrs, autism and DD)	Non-compliant vocal behaviour ***	ABC	1)Directive prompts (e.g., "show me the X") Vs. 2) Non-directive prompts (e.g., "I wonder where the X is").	Multi-element	Standard 1-Y Standard 2-Y Standard 3- N Standard 4- R	Reduction in problem behaviour in non-directive prompting conditions. No influence on compliance
Borrero, Vollmer and Borrero (2004b)	*Tobias* (13yrs, moderate ID)	Aggression	ABC	1)Abrasive Vs. 2)Neutral prompts during demands	Pairwise multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4- R	Problem behaviour more likely following abrasive prompts

Northup, Kodak, Lee, & Coyne, (2004)	*Marie* (5yrs, ADHD)	Inappropriate behaviour	ABC	Manipulated description of contingency. Phase 2 =1) "Taking a break" Vs. 2) "Time out". Phase 3= 1) No description Vs. 2) "Time out"	Multi-element	Standard 1-Y Standard 2-Y Standard 3- Y Standard 4- R (phase 2), Y (phase 3)	Inappropriate behaviour at near zero rates when escape contingency verbally described as "time out".
Stichter, Sasso & Jolivette (2004)	*Josh* (7 yrs, emotional and behavioural difficulties)	Aberrant behaviour Off-task behaviour	Antecedent	Phases 1-3 manipulated combinations of: 1) background noise, 2) social contact, 3) task structure. Phase 4: 1)High structure Vs.2) Control..	Phase 1: non-experimental ABCDEF Phase 2: ABA Phase 3: ABCDED Phase 4: ABAB	Standard 1-N[12] Standard 2-Y Standard 3- Y (phase 4 only) Standard 4- R (phase 4 only)	High structure reduced levels of off-task behaviour. No clear effect on levels of aberrant behaviour.
Carr, Smith, Giacin, Whelan, & Pancari (2003)	5 males (age 30-48 yrs, autism and mild-profound ID) 3 females (age 29-48yrs, 1	Problem behaviour ***	Antecedent	1) Good mood, plus demands Vs. 2) Good mood, no demands Vs. 3) Neutral mood, plus demands Vs.4) Neutral mood, no demands Vs. 5) Bad	Multi-element manipulation of demand Vs. no demand conditions embedded within naturally occurring	Standard 1-N Standard 2-Y Standard 3-Y Standard 4-Y	All participants more sessions terminated due to problem behaviour in demand probe when participants rated as being in a

[12] No measure of treatment integrity despite using peer as 'change agent' in natural settings

	with autism, moderate-profound ID)			mood, demands Vs. 6)Bad mood, no demands	variation in 'mood'		'bad mood'
Ebanks & Fisher (2003)	J*im*, (19 yrs, ID and pervasive DD)	Destructive behaviour	ABC	1)Standard corrective prompting procedure Vs. 2) Antecedent prompting procedure.	ABAB reversal	Standard 1-Y Standard 2-Y Standard 3- Y Standard 4- R	Destructive behaviour at zero rates in antecedent prompting
Asmus et al (1999)	*Luke* (3 yrs, moderate ID) *Todd* (4 yrs, XYY syndrome, moderate ID) Trevor (5 yrs, moderate-severe ID, ADHD)	Aberrant behaviour ***	Antecedent (all participants) ABC (Todd, Trevor)	Phase 1:1). Task instruction Vs.2) Free play Phase 2: Effects of manipulating familarity of 1) task instructions, 2) therapist, 3) setting.	Combination multi-element and reversal.	Standard 1-Y Standard 2-Y Standard 3- Y Standard 4- R	Presence of task instructions, not materials/setting or therapist associated with aberrant behaviour. Unfamiliar tasks quickly acquired aversive properties and evoked aberrant behaviour.

Task Difficulty/Preference

Task difficulty has been long-recognized as an important variable influencing the aversiveness of demands (Carr & Durand, 1985; Weeks & Gaylord-Ross, 1981). The review identified several studies to support this position (see Table 6). A range of methods were used to help identify 'difficult' demands; including staff report (Butler & Luiselli, 2007), classroom approach behaviours (Reichle, Johnson, Monn, & Harris, 2010), the use of a demand hierarchy assessment (Call, Wacker, Ringdahl, Cooper-Brown, & Boelter, 2004), and task accuracy (Lee, Sugai, & Horner, 1999; Moore & Edwards, 2003; Reichle & McComas, 2004). All these studies demonstrated that demands rated as 'difficult' were more likely to evoke escape-maintained problem behaviour than demands rated as 'easy'.

A number of studies demonstrated the utility of interventions designed at reducing the difficulty of a task *either* by altering supports available to the individual *or* by teaching adaptive behaviours to help the individual complete the task. McComas, Hoch, Paone and El-Roy (2000) showed that the introduction of instructional strategies to reduce task difficulty (use of calculator, number lines) successfully reduced the occurrence of escape-maintained problem behaviour for a boy with autism. Both Lee, Sugai and Horner (1999) and Lalli, Kates and Casey (1999) identified the absence of component skills served to make specific tasks difficult and thereby become associated with escape-maintained problem behaviours. Both studies reported on instructional interventions designed at teaching students the component skills required to complete a task successfully and thereby reduce its difficulty and the probability of escape-maintained problem behaviour.

Such findings have important implications for both the assessment and treatment of problem behaviour. In relation to the assessment of problem behaviour, it seems important that task difficulty should be assessed prior to the selection of tasks for the demand condition of a functional analysis. There is a wealth of data to suggest that failure to include tasks of sufficient difficulty will fail to evoke the same level of problem behaviour than would otherwise be expected. If task difficulty is found to be an important variable then clinicians should find opportunities to either: a) alter the task to reduce its difficulty, b) increase the level of support and c) ensure that the student has the full repertoire of skills required to complete the task.

Table 6. *Studies that Demonstrate the Influence of Alterations to Parameters of Task Difficulty/Preference as an MO for Negatively Reinforced Problem Behaviour.*

Study	**Participants (age, diagnoses)**	**DV**	**Functional assessment**	**IV**	**Design**	**Experimental standards met**	**Findings**
Reichle, Johnson, Monn, & Harris, (2010)*	*Mark* (4 yrs, autism, severe cognitive delay) *Pete* (4 yrs, ASD)	Challenging behaviours***	Antecedent	1) Tasks associated with approach Vs. 2) Tasks never approached	Alternating treatments design	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Lower rates of problem behaviour and more engagement in tasks associated with approach
Butler & Luiselli, (2007)*	*Michelle* (13yrs, autism)	Problem behaviour	ABC	Comparison of three requests of differing difficulty	Multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-N	Difficult task associated with high rates of problem behaviour.
Call, Wacker, Ringdahl, Cooper-Brown, & Boelter, (2004) *	*Daisy* (4yrs) *Andy* (6 yrs) *Zach* (8yrs) *Jacob* (5 yrs)	Non-compliance	ABC	1) Decreased amount of demands Vs. 2) Decreased difficulty (Zach, Jacob, and Andy) Vs. 3) Decreased difficulty of demands plus attention (Daisy and Jacob).	Brief multi-element design	Standard 1-Y Standard 2-Y Standard 3-N Standard 4-N	Decreasing difficulty and amount reduced non-compliance for all except Andy.

Reichle & McComas (2004)	*Timothy* (12yrs, cognitive delay)	Challenging behaviour	Antecedent	1) 'Easy' task Vs. 2) 'Difficult' task	ABAB reversal using single probes	Standard 1-Y Standard 2-N Standard 3-Y Standard 4-N	Higher levels of problem behaviour in 'difficult' task condition
Moore & Edwards (2003)*	*Jacob* (17 yrs) *Robert* (7 yrs) *Edgar* (9yrs) Morris (7 yrs)	Problem behaviour	ABC	1) Easy task/high attention Vs. 2) Easy task/low attention Vs.3) Hard task/low attention Vs. 4) Hard task/high attention	Multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-N	Task difficulty primarily related to problem behaviour for both Robert and Jacob.
McComas, Hoch, Paone, & El-Roy (2000)	*Eli* (8yrs, autism, DD)	Destructive behaviour	ABC	1) Instructional strategy (e.g., calculator, number lines) Vs. 2) No strategy	Multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	More problem behaviour and lower compliance in conditions without instructional strategy
Lee, Sugai, & Horner (1999)	*Bill* (9 yrs, specific learning difficulties) *Matt* (9 yrs, ADHD)	Problem behaviour Off task behaviour *	Descriptive Antecedent	Comparison of easy Vs. difficult tasks	ABA reversal	Standard 1-Y Standard 2-Y Standard 3-N Standard 4-Y	Higher problem behaviour and lower compliance during difficult tasks. Intervention focused on teaching component skills

							reduced problem behaviour.
Lalli, Kates & Casey (1999)	*Jake* (11yrs, mild ID) *Mark* (10 yrs, mild ID)**	Aggression*	Brief ABC	Compared different task associated with problem behaviour.	Brief probe multi-element	Standard 1-Y Standard 2-Y Standard 3-N Standard 4-N	Spelling evoked aggression. Skills teaching improved accuracy/ aggression

Choice/Predictability of Tasks

As reported in McGill's (1999) review, manipulations to both choice (Dunlap, Kern-Dunlap, Clarke, & Robbins, 1991; Foster-Johnson, Ferro, & Dunlap, 1994) and the predictability of tasks (Flannery & Horner, 1994) have been shown to reduce the occurrence of escape-maintained problem behaviour. This literature has continued to grow in recent years (see Table 7). With regard to task predictability, Reichle, Johnson, Monn, and Harris (2010) demonstrated that the use of explicit cues signalling the end of a task (such as "only X more to go") resulted in reductions in the escape-maintained problem behaviour of two four year old boys with an autistic spectrum disorder, in comparison to the use of more general delay cues (e.g., "only a few left"). Studies that have examined choice have shown that offering choice over the sequence of tasks, the type of reinforcers available and having the option to select a different task once instruction has begun can reduce escape-maintained problem behaviour (McComas, Hoch, Paone, & El-Roy, 2000; Newman, Needelman, Reinecke, & Robek, 2002; Romaniuk et al., 2002). Romaniuk et al. (2002) demonstrated that a choice-making intervention reduced the escape-maintained problem behaviour of four participants but had no influence on attention-maintained problem behaviour, demonstrating the importance of matching an intervention to behavioural function. Part of the choice-making strategy employed by Romaniuk et al. involved offering the opportunity to choose a change of tasks during instruction. Findings by McComas, Hoch, Paone, and El-Roy (2000) suggest that, for one participant at least, task repetition may have aversive properties and one effect of choice-making may be to interrupt the aversiveness of task repetition.

Table 7. *Studies that Demonstrate the Influence of Altering the Level of Predictability, Control or Choice on Negatively Reinforced Problem Behaviour.*

Study	**Participants (age, diagnoses)**	**DV**	**Functional assessment**	**IV**	**Design**	**Experimental standards met**	**Findings**
Reichle, Johnson, Monn, & Harris (2010) *	*Mark* (4 yrs, autism, severe cognitive delay) *Pete* (4 yrs, ASD)	Challenging behaviour***	Descriptive	1)Explicit Vs. 2) General delay cue	Alternating treatments design with changing criterion	Standard 1-N[13] Standard 2-Y Standard 3-Y Standard 4-Y	Greater reductions in problem behaviour and increases in compliance following explicit delay cues.
Newman, Needelman, Reinecke, & Robek (2002)	3 males with autism. (Andy, Carl, Paul) Age range 7-12 yrs	Competing behaviour ***	Indirect	1) Teacher selection of reinforcers and task order Vs. 2) Student selection	Alternating treatment design	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Higher rates of competing behaviours occurred in teacher selection conditions than student selected. No impact on accuracy.
Romaniuk et al (2002)	*Brooke* (7 yrs, Cerebal Palsy, moderate cognitive impairments)	Problem behaviour	ABC	1)Choice Vs. 2) No choice	Reversal; ABAB (Brooke,	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Lower problem behaviour occurring in the choice condition. Unlike 3

[13] Alternating treatments design but two different tasks and different criterion between two conditions means not comparing like with like

	Maggie (10 yrs, autistic disorder, IQ score in low-average range)Gary (7yrs, moderate ID) *Katie* (8 yrs, mood disorder unspecified, moderate cog impairment) **				Gary) ABABAB (Maggie) Multi-element (Katie)	(Brooke, Gary, Katie), N (Maggie)	participants with attention-maintained behaviour. Katie showed a reduction in escape-maintained but not attention-maintained behaviour .
McComas, Hoch, Paone, & El-Roy, (2000) *	*Charlie* (8 yrs, DD, autism) *Ben* (9 yrs, DD, autism)	Aggression, SIB ***	ABC	Charlie: Compared: 1) choice over sequence of tasks, 2) no choice. Ben: Compared: 1) Non-repeated task (given worksheets not previously completed), 2) Repeated task (given worksheets already completed)	Multi-element	Standard 1-Y Standard 2:Y Standard 3-Y Standard 4-Y	Charlie: Some evidence of differentiation for SIB, however DRO added to reduce SIB to acceptable levels. No influence on compliance. Ben: higher aggression in repeated task condition. No difference for compliance.

Non-contingent Escape and Embedding Demands in Contexts Associated with Positive Reinforcement

McGill (1999) provided a MO interpretation of non-contingent escape (NCE) as an intervention for problem behaviour and studies to have examined this intervention were cited in his paper (e.g., Vollmer, Marcus, & Ringdahl, 1995). The review identified a number of studies that demonstrated the use of providing breaks from aversive demands on a fixed-time schedule (see Table 8). These studies have shown variants of this intervention to be effective in reducing the escape-maintained behaviour of individuals with intellectual and developmental disabilities (Aikman, Garbutt, & Furniss, 2003; Kodak, Miltenberger, & Romaniuk, 2003b; Wesolowski, Zencius, & Rodriguez, 1999), typically developing children undergoing dental treatment (O'Callaghan, Allen, Powell, & Salama, 2006), and an older adult with Alzheimer's disease (Baker, Hanley, & Mathews, 2006).

In his review, McGill (1999) noted that embedding a demand within a context containing preferred events or activities, such as storytelling (Carr, Newsom, & Binkoff, 1976), social comments (Kennedy, Itkonen, & Linquist, 1995) or following *high p* demands[14] (Mace & Belfiore, 1990) could reduce the occurrence of escape-maintained problem behaviour.

This important area of research has continued to attract attention in the literature (see Table 9). Research has continued to demonstrate the positive influence of preceding a *low-p* request with a request that is highly likely to be complied with for problem behaviours maintained by escape (Patel et al., 2006). A number of studies have demonstrated that presenting

[14] Involving the presentation of a demand assocaited with a high probability of compliance prior to the presentation of a demand with low probability of compliance

demands within the context of an ongoing preferred activity may reduce the occurrence of escape-maintained problem behaviour (Carey & Halle, 2002; Wilder, Normand, & Atwell, 2005). Similarly, manipulations made to the quality of attention available during demands (Call, Wacker, Ringdahl, Cooper-Brown, & Boelter, 2004; Gardner, Wacker, & Boelter, 2009) or preference for the specific materials (e.g., toys; Boelter et al., 2007; Harding et al., 1999) used in demand procedures have been shown to influence escape-maintained problem behaviours. Harding et al. (1999) adopted a concurrent choice procedure to examine the influence such manipulations had on problem behaviour and response allocation. Interestingly, one participant allocated her responses to conditions in which the manipulation of a highly preferred toy was used for instructional activities and this was associated with higher task completion and lower levels of problem behaviour. However, for another participant a similar manipulation increased rates of problem behaviour and reduced task completion, suggesting it was aversive. Indeed, for some individuals restricting access to preferred items during demand presentation has been shown to evoke escape-maintained problem behaviours (Call, Wacker, Ringdahl, & Boelter, 2005). It may be that the restricted nature of interactions with the highly preferred toy in the Harding et al. study served to establish this manipulation as aversive.

Other studies have examined providing non-contingent access to attention or tangibles on either a fixed interval or variable interval schedule (Ingvarsson, Hanley, & Welter, 2009; Ingvarsson, Kahng, & Hausman, 2008; Lomas, Fisher, & Kelley, 2010; Long, Hagopian, DeLeon, Marhefka, & Resau, 2005; Reed et al., 2004). These studies have reported somewhat mixed results. For example, Lomas, Fisher and Kelley (2010) reported that providing access to praise and food on a variable interval schedule reduced the problem behaviour of three

participants with Autistic Spectrum Disorder but only improved the compliance of one participant. Whilst Ingvarsson et al. in two related studies have noted the benefits of providing non-contingent access to food tangibles, they reported that the density of NCR had little impact on compliance or problem behaviour (Ingvarsson, Kahng, & Hausman, 2008); they also reported minimal difference between NCR and providing reinforcement contingent on compliance (Ingvarsson, Hanley, & Welter, 2009). Reed et al. (2004) reported the NCR only reduced problem behaviour when combined with escape extinction, suggesting that for some individuals at least, NCR may not be sufficient to achieve behavioural change.

This body of research suggests that the aversiveness of a demand may be altered by introducing potential sources of positive reinforcement either prior or during the onset of an activity. Whilst such a manipulation may have unintended consequences for some children (e.g., Harding et al., 1999), this form of intervention may be an important consideration in applied contexts. This may be especially important to consider in situations where an individual is considered to be in a 'bad mood' and may be more likely to present with problem behaviours following a demand (Carr, McLaughlin, Giacobbe-Grieco, & Smith, 2003). Such an approach may represent a specific example of more general notion of "neutralising" (Horner, Day, & Day, 1997) the effects of pre-existing MOs. In relation to assessment, it seems important that the quality of attention provided during demands and preference for task materials is controlled. Studies appear to show that providing access to 'high quality' attention or to highly preferred materials may alter the probability of escape maintained problem behaviour. Likewise, it is possible that providing access to highly preferred materials during demand activities could restrict the way in which a child interacts with them and could thereby evoke tangible-maintained

problem behaviour. Within-session analyses would be an important means of examining such relationships where they are suspected to exist.

Table 8. *Studies that Demonstrate the Influence of Altering the Schedule of Demands as an MO for Negatively Reinforced Problem Behaviour.*

Study	**Participants (age, diagnoses)**	**DV**	**Functional assessment**	**IV**	**Design**	**Experimental standards met**	**Findings**
Baker, Hanley, & Mathews (2006)* Phase 3	*Participant* (96 yrs, Alzheimer's dementia)	Aggression	ABC	1)Non-contingent escape (NCE) Vs. 2) Contingent escape	ABA	Standard 1-Y Standard 2-N Standard 3-N Standard 4-Y	Problem behaviour at lower rates in the NCE condition
O'Callaghan, Allen, Powell, & Salama, (2006)	Five typically developing children, ranging from 4 to 7 years old. 3 girls (Melissa, Tanya, Elaine) and 2 boys (George,	Disruptive behaviour	Topography	1) Routine restorative dental treatment and 2) NCE	Multiple baseline design (across participants)	Standard 1-N[15] Standard 2-Y Standard 3-Y Standard 4-Y	Four children showed reductions following the introduction of NCE.

[15] IV is different across each 3 minute session. Different procedures (and MOs) according to different visits. Unclear whether change is due to IV or confound (i.e., procedures different within and between each visit).

	Kevin)						
Aikman Garbutt, & Furniss (2003)	*Beth* (8yrs, severe DD)	Screaming and throwing	Descriptive	1)Continuous demands Vs. 2) Interspersed 5min demand with 5min play.	Brief multi-element probe	Standard 1-N[16] Standard 2-N Standard 3-N Standard 4-N	Higher levels of throwing and screaming in 'high' continuous demand condition Vs. intervention condition.
Kodak, Miltenberger, & Romaniuk (2003a)	*Andy* (4yrs, autism) *John* (4yrs, autism)	Disruptive behaviour ***	Unclear	1) NCE Vs. 2)DNRO	Multiple-baseline across participants with embedded alternating treatment design.	Standard 1-Y Standard 2-Y Standard 3-N Standard 4-N	NCE led to reductions in escape maintained problem behaviour and increases that were similar in magnitude to DNRO
Wesolowsk, Zencius, & Rodriguez (1999)	*Jim* (19yrs, frontal lobe TBI). *Ralph* (16 yrs, frontal lobe TBI). *Mark* (24yrs, frontal lobe TBI)	Frequency of unauthorised breaks	Topography	1)Baseline: 2x15 minute breaks/day Vs. 2)5x 10 minute breaks/day	Multiple-baseline across participants	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Introduction of regular schedule of breaks reduced unauthorised breaks of all three participants.

[16] Teacher as interventionist but no fidelity measure

Table 9. *Studies that Demonstrate the Influence of Adding Potential Sources of Positive Reinforcement to the Demand Context as a MO for Negatively Reinforced Problem Behaviour.*

Study	**Participants**	**DV**	**Functional assessment**	**IV**	**Design**	**Experimental standards met**	**Findings**
Lomas, Fisher, & Kelley (2010)	*Sam* (8yrs Asperger, ADHD, mild DD) *Aaron* (8yrs, autism) *Mark* (9yrs, autism)	Problem behaviour ***	ABC	1)Demand baseline Vs. 2)Praise and edible tangible on VT 15s schedule	Reversal ABAB	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Reduction in problem behaviour for all participants. Compliance increased for Aaron only
Ingvarsson, Hanley, & Welter (2009)	*Erika* (3 yrs) *Mark* (3 yrs) *Jason* (3 yrs)	Disruptive behaviour, Problem behaviour ***	ABC	1)Contingent reinforcement Vs.2) Non-contingent reinforcement (NCR)	Erika (alternating treatments design) Jason, Mark ABA with embedded alternating treatment design	Standard 1-Y Standard 2-Y Standard 3-N Standard 4-Y	NCR effective in reducing problem behaviour for two participants (Erika and Mark) and increasing compliance

Gardner, Wacker, & Boelter, (2009)	*Kurtis* (8yrs) *Carter* (6yrs)	Inappropriate behaviour	ABC	1)Demand with low quality attention Vs. 2)Demand with high quality attention. Compared within functional analysis	Brief multi-element design	Standard 1-N Standard 2-Y Standard 3-N Standard 4-N	Inappropriate behaviour at higher levels in low quality attention demand condition.
Ingvarsson, Kahng, & Hausman (2008)	*Manuela* (8 yrs, autism, moderate ID)	Problem behaviour ***	ABC	1)Demand baseline Vs. 2) NCR (food tangibles) delivered at both high density and low density	ABAB (with embedded alternating treatments design)	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	NCR reduced problem behaviour and increased compliance in comparison to baseline. Minimal difference between high or low density
Boelter et al (2007)	*James* (4 yrs, ADHD)	Disruptive behaviour***	Indirect	1)Demand + high preference toys Vs. 2) Demand +low preference toys	Multi-element probe design with reversals	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-N	Higher rates of disruptive behaviour and lower accuracy in demand conditions that involved manipulating low preference toy

Patel et al (2006)	*Kisha* (2 yrs, DD) *Simone* (2 yrs, mild DD)	Inappropriate behaviour **	Topography	1) Extinction + preceding low p demand with high p demand Vs. 2) escape extinction on its own	Kisha (Reversal design, ABABA) Simone (alternating treatment design embedded in ABAB reversal design)	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Lower rates of inappropriate behaviour and higher rates of food acceptance occurred for Kisha and Simone under high p conditions
Call, Wacker, Ringdahl, & Boelter (2005)	*Kevin* (2yrs)	Aggression	ABC	Combined demand + restricted tangible. condition as part of functional analysis.	Multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Higher rate of problem behaviour in combined condition.
Long, Hagopian, DeLeon, Marhefka and Resau (2005)	*Trent* (9yrs, autism, severe ID) *Marsha* (19yrs, profound ID) *Janelle* (5yrs, profound ID)	SIB, aggression, disruption	ABC	1) Competing stimuli Vs. 2) Baseline during hygiene-related demands	ABAB	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Problem behaviours reduced during presentation of competing stimuli
Wilder, Normand, & Atwell (2005)	*Raley* (3yrs, autism)	SIB ***	Brief ABC	1)Baseline escape Vs. 2) Continuous access to preferred video	ABAB design	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-R	Lower levels of SIB and higher levels of acceptance in video conditions

Reed et al (2004)	*Jansen* (1 yr) *Nate* (1yr) *Jaden* (3yrs, DD) *Abbott* (4yrs, Pierre Robin syndrome)	Inappropriate behaviour, Negative vocalisations	Topography	A phase:1) Escape baseline Vs. 2) NCR with escape B phase: 1) Escape extinction Vs. 2) NCR with escape extinction	Multi-element design embedded within an ABAB reversal	Standard 1-Y Standard 2-Y [except Abbott drinking data] Standard 3-Y Standard 4-R	NCR alone did not lead to decreases in inappropriate behaviour.
Call, Wacker, Ringdahl, Cooper-Brown, & Boelter (2004)* Phase 2	*Andy* (6 yrs) *Zach* (8yrs) *Jacob* (5 yrs)	Non-compliance (off task behaviour and problem behaviour)	ABC	Effects of providing attention concurrent with demands during functional analysis	Brief multi-element design	Standard 1-Y Standard 2-Y Standard 3-N Standard 4-N	Lower rates of problem behaviour in demand + attention condition for Zach only.
Carey & Halle (2002)	*Steve* (12yrs, severe ID)**	SIB	ABC	Embedding demands in context of preferred activity (listening to music)	Multi-element	Standard 1-Y Standard 2-Y Standard 3-Y Standard 4-Y	Lower escape-maintained behaviour when demands presented with music.
Harding et al (1999)	*Susan* (4 yrs, autism and DD) *Kyle* (4 yrs, behaviour disorder)	Problem behaviour ***	ABC	Both: [A] Alone with low preference toy Vs. Instruction with high preference toy. [B] Alone with high preference toy Vs.	Concurrent schedules combined with reversal design Susan (ABABA)	Standard 1-Y Standard 2-Y Standard 3-Y (Susan), N (Kyle) Standard 4-R (Kyle,	Susan: Choice A bias towards instruction with toy. Choice B bias toward area that allowed escape from instructions while gaining

	instruction with low preference toy) Kyle only:[C] Alone with high preference toy Vs. instruction with high preference toy)	Kyle (ABACB)	Susan)	access to the highly preferred toy. Kyle: Avoided instructions associated with a highly preferred toy (in comparison to low preference toy) and was more likely to display problem behaviour under such conditions.

Conclusion

Over the past decade there have been considerable developments in the investigation of MOs and in the role played by such variables in problem behaviour maintained by negative reinforcement. 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 problem behaviour and has been beneficial in developing the understanding of negatively reinforced problem behaviour. The implications of these developments for the assessment, treatment and study of negatively reinforced problem behaviour have been outlined throughout this review.

The findings of the review highlight the importance of attending to person characteristics in their environmental context and to the interplay between the two (see Figure 1). The review has highlighted a number of environmental and person-level variables that could be considered to act as 'risk markers' for negatively reinforced problem behaviour. The risk of negatively reinforced problem behaviour will be elevated in certain environmental contexts (for example, environments in which aversive stimuli are not embedded in a 'positive' context, environments lacking opportunities for choice or control)[17]. Likewise, there appears to be certain person-level variables that are associated with a heightened propensity to display negatively reinforced problem behaviour under certain environmental conditions (for example, the presence of specific phenotypes associated with genetic syndromes and health conditions or the absence of certain behavioural repertoires). Whilst ameliorating environmental MOs may be sufficient to reduce

[17] It should be noted that there are idiosyncratic responses to such manipulations across different individuals.

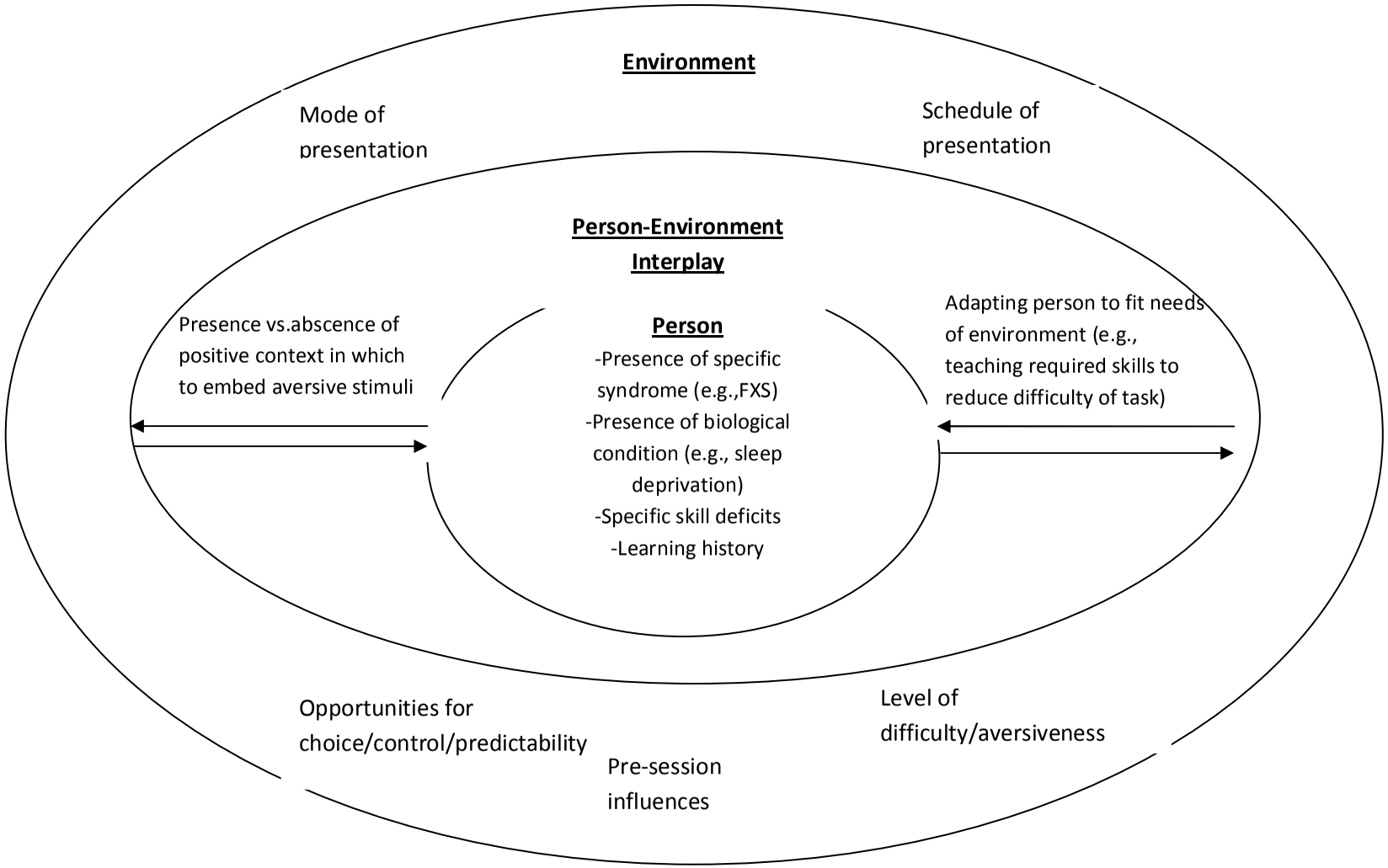

Figure 1. Person-Environment Interplay and Motivating Operations for Negatively Reinforced Problem Behaviour.

negatively reinforced problem behaviour for many individuals, in cases where person-level variables play a role then their interplay with environmental factors will need to be targeted in treatment (Carr & Smith, 1995). Such interventions may take the form of adapting the environment in order to meet the needs of the individual (for example, by reducing the presentation of specific aversive stimuli, such as eye contact in Fragile X syndrome) or attenuating or accentuating person characteristics to meet the needs of the environment (for example, using ear plugs to reduce the impact of noise in Williams syndrome or teaching specific behavioural repertoires to the individual).

As reflected in this review, the behaviour analytic literature on problem behaviour has shown a bias towards the assessment of environmental contributions to problem behaviour. This understanding can only be strengthened and extended by considering more person-level sources of variability. The results of this review suggest that behavioural assessment (and intervention) based solely on a functional assessment of environmentally-based variables, whilst necessary, may be an incomplete account of problem behaviour. The incorporation of traditional behaviour analytic models of functional assessment with an approach that also considers the characteristics and needs of the person (Emerson & McGill, 1989; LaVigna & Willis, 2005) would therefore appear to be beneficial in extending existing accounts.

As has been highlighted, there are several areas where future research is needed in order to continue this advancement. At present, current understanding of the role of a number of important variables (i.e., role of biological variables, influence of pre-session events) is incomplete and a more systematic approach towards research on the role of MOs in negatively reinforced problem behaviour would be beneficial. The developments that have taken place over

the past decade, however, 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 negatively reinforced problem behaviour.

PREFACE TO CHAPTER 2

The literature review has highlighted the importance of person-environment interactions for problem behaviours maintained by negative reinforcement. Phenotypic variation related to specific genetic syndromes represents a salient 'person-level' variable. Very few studies have investigated the effects of adapting the environment to ameliorate the difficulties posed by phenotypic influences. In one of the few examples of this important line of research, O'Reilly et al. (2000) demonstrated that the use of ear plugs served to neutralise the effects of hyperacusis in a child with Williams syndrome and reduced both pain-related and escape-maintained problem behaviours.

The empirical paper presented in the next chapter attempted to further this line of research by examining the influence of eye contact on behaviour in children with fragile X syndrome (FXS). Gaze avoidance is a central feature of FXS. Whilst the function served by such behaviours remains poorly understood, it has been suggested that people with FXS find eye contact aversive. This phenotype was therefore selected as an exemplar with which to investigate the types of person-environment interaction described in Chapter 1. The gaze-avoidance and arousal-related behaviours of four boys with FXS were investigated under conditions of high and low eye contact, using single case experimental design. Between-session and within-session analyses were conducted in order to help delineate the relationship between such manipulations and gaze- and arousal-related variables.

CHAPTER 2: EMPIRICAL PAPER

EMPIRICAL EXAMINATION OF THE INFLUENCE OF EYE CONTACT ON GAZE AVOIDANCE IN FRAGILE X SYNDROME

Abstract

Gaze avoidance is a central feature of the behavioural phenotype of fragile X syndrome. However, the function served by such gaze avoiding behaviours remains poorly understood, and has been typically inferred by topography. The current study employed single-case experimental design methodology to further delineate the function served by such behaviours. Variation in gaze avoidance and arousal-related behaviours were examined under conditions of high- and low-eye contact in four boys with a diagnosis of fragile X syndrome. Findings indicated that specific topographies of gaze-avoidance behaviour were more likely to occur under high-eye contact conditions for each participant. Within-session lag sequential analyses suggested that the occurrence of these behaviours was consistent with a negative reinforcement hypothesis for three participants. Evidence is also presented to suggest an association between gaze avoidance behaviours and increases in arousal. These findings are related to the existing literature on gaze avoidance in fragile X syndrome and a model to account for such findings in FXS is postulated [18].

[18] The paper has been prepared as if for submission to the *American Journal on Intellectual and Developmental Disabilities.*

Introduction

Fragile X syndrome (FXS) is the primary inherited cause of intellectual and developmental disability. FXS is caused by an increase in the number of trinucleuotide repeats of cytosine-guanine-guanine (CGG) within the FMR1 gene (Verkerk et al., 1991). For individuals with the full FMR1 mutation, the number of CGG repeats exceeds 200, resulting in the hypermethylation of the promoter region of the FMR1 gene. This restricts the production of the FMR1 protein product (FMRP), which plays an important role in the regulation of brain proteins (Lightbody & Reiss, 2009; Bagni & Greenough, 2005). As the genetic locus of FXS is located on the X chromosome, males are typically more adversely affected than are females. FXS is associated with mild to profound intellectual and developmental disability and a range of physical, cognitive, and behavioural sequalae (Cornish, Turk, & Hagerman, 2008).

One striking behavioural feature of FXS is the high levels of gaze aversion. Questionnaire studies reported that, in comparison to contrast groups, individuals with fragile X syndrome show greater problems with establishing eye contact (Einfeld, Tonge, & Florio, 1994; Einfeld, Tonge, & Turner, 1999; Lachiewicz, Spiridigliozzi, Gullion, Ransford, & Rao, 1994; Meerenstein et al., 1996). Meerenstein et al. (1996) reported that 80-88% of males with fragile X syndrome showed poor eye contact. A seven year longitudinal study noted that gaze aversion (together with shyness) distinguished children with FXS from controls at both baseline and follow up (Einfeld et al., 1999).

Studies adopting direct behavioural measures have reported within- and between-group variations in eye contact within FXS (Cohen et al., 1988; Farzin, Rivera, & Hessl, 2009; Garrett, Menon, MacKenzie, & Reiss, 2004; Hagerman et al., 1992; Wolff, Gardner, Paccia, & Lappen,

1989). Wolff et al. (1989) examined the greeting behaviour of 18 males with FXS, reporting a characteristic profile of gaze avoidance (involving a whole body turn) in comparison to a group of 18 males with Down syndrome. Farzin, Rivera and Hessl (2009) demonstrated that children with FXS made fewer fixations to the eye region of facial stimuli and spent less overall time looking at this area than did sex and age-matched typically developing controls.

Whilst gaze aversion has been widely reported in children with autism (Senju & Johnson, 2009a); it appears that children with FXS show a different pattern of gaze avoidance than do children with autism. Cohen et al (1988) compared children with and without FXS, who also had a diagnosis of autism or were classified as atypically developing. Whilst boys with FXS showed a higher overall level of gaze avoidance across all conditions, they were, unlike children with autism, able to discriminate between strangers and parents (i.e., there was less avoidance shown to parents relative to strangers). Studies also suggest that familiarity exerts an abative influence on gaze avoidance in FXS (Cooper, 1993; Hall, Lightbody, Huffman, Lazzeroni, & Reiss, 2009; Roberts, Weisenfeld, Hatton, Heath, & Kaufmann, 2007). For example, Hall et al. (2009) reported small but significant reductions in the gaze avoidance of males with FXS, but not their typically developing peers, over the course of a 25-minute social interaction. Roberts et al (2007) reported children with FXS showed improvements in social approach behaviours as the amount of time with the experimenter increased. In a related study, Roberts et al (2009) found evidence of a 'warm up' effect for boys with FXS but not for those who also met criteria for autism (scoring >30 on the CARS). As such it appears that there is variability in the degree of gaze aversion that children with FXS present with and that this may in part be a function of familiarity and autism spectrum disorder.

Researchers have primarily adopted a 'hyperarousal' hypothesis to account for gaze avoidance in FXS (Belser & Sudhalter, 1995; Hall, Lightbody, Huffman, Lazzeroni, & Reiss, 2009; Hessl et al., 2002; Hessl, Glaser, Dyer-Friedman, & Reiss, 2006). According to this hypothesis the social demands of eye contact when paired with heightened anxiety, lead to hyperarousal, which the person with FXS seeks to avoid. This hypothesis argues therefore that eye contact is, at least in part, aversive for children with FXS due to its association with social demands. There is some evidence to support this hypothesis. Cohen et al (1989) compared 24 males with FXS against 24 males with autism and eight typically developing mental-age matched controls during a 10-minute interaction between parent and child. Lag sequential analyses demonstrated that whilst males with FXS responded to the initiation of eye contact by their parents, they found such contact aversive (i.e., engaged in escape-behaviours). Males with autism showed an opposite pattern of results (i.e., were not sensitive to parental initiation of eye contact but did not find such contact aversive). These findings were taken to support the hypothesis that males with FXS show selective avoidance of mutual gaze relative to social gaze, but remain sensitive to gaze initiation by parents. A preliminary study by Belser and Sudhalter (1995) reported that two participants with FXS showed higher skin conductance level and language errors in conditions characterized by higher levels of eye contact during a 5-10 minute conversation. However, the study was confounded with the eye contact condition always occurring first. Hall, DeBernardis, and Reiss (2006) reported that social escape behaviours were more likely to occur in conditions with high social demands and higher levels of eye rubbing, hand biting and hyperactivity were found in an 'Interview' condition, which also included repeated prompts for eye contact.

Studies have also examined the relationship between cortisol, a marker of the activation of the L-HPA axis (Lopez, Vazquez, & Olson, 2004), and gaze aversion. In their study, Hall et al (2006) noted that children with FXS with higher mean levels of cortisol showed lower levels of eye contact. Hessl et al (2006) reported that for males with FXS the quality of eye contact with an unfamiliar adult was strongly correlated with cortisol reactivity. Somewhat paradoxically, children with FXS who had the highest levels of gaze avoidance were found to have the lowest levels of cortisol reactivity in response to the social challenge task. These contrasting findings may reflect the effectiveness of gaze aversion in reducing the aversiveness of the situation (and hence the level of arousal experienced by the child). It may be that children with higher mean levels of cortisol (as shown in the Hall study) are more likely to engage in escape or avoidance behaviours in response to social demands, resulting in a lowering of cortisol levels following the presentation of a stressor (as found in the Hessl study), which would be consistent with a negative reinforcement account of gaze aversion.

One difficulty with all of these studies is that the social and information processing demands of tasks have been confounded. This has made it difficult to determine whether eye gaze *per se* is aversive or whether it is the task demands (independent of eye contact) that are aversive and evoke gaze avoidance as an operant response. Some evidence for this latter hypothesis has been provided by Murphy et al (2007). Murphy evaluated gaze avoidance in both a 'Social' condition (in which an experimenter sat in front of the participant whilst a task was presented via a computer) and a 'Non-social' condition (in which the experimenter sat behind the participant whilst completing the same task). Regardless of either task difficulty or the positioning of the experimenter, participants with FXS showed greater levels of gaze avoidance than did typically

developing controls or those with Down syndrome. There was no difference in gaze avoidance between either the Social or Non-social conditions, indicating that 'social' aspects of the demand were not responsible for the gaze avoidance. Similarly, variations in task difficulty did not influence gaze avoidance in FXS but did in other groups. Finally, there was no evidence to indicate that a combination of high task *and* high social demands were associated with gaze avoidance. Therefore, according to these authors gaze avoidance in FXS may simply reflect a generalised negatively reinforced behaviour that serves to escape or avoid aversive situations or contexts, rather than being necessarily related to eye contact per se[19].

The current study employed an empirical examination to: determine whether eye contact functions as an aversive stimulus for children with fragile X syndrome and further delineate the nature of the relationship between eye contact and gaze avoidance behaviours in boys with fragile X syndrome.

Specifically, the aim of the study was to elucidate the function served by gaze avoidant behaviours by examining whether: 1) systematic manipulations made to the level of eye contact during instructional demands influenced the occurrence of gaze avoidance behaviours; 2) different topographies of gaze avoidance were related within response class hierarchies; 3) gaze avoidance behaviours were evoked by prompts for eye contact and/or the onset of task-related

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[19] *"There may be nothing about eye contact per se that is arousing; rather, it may be the sensory stimulation and/or high order demands associated with eye contact that is problematic for individuals with fragile X syndrome (Murphy et al, 2008; p. 356)"*

demands and 4) gaze avoidance behaviours were associated with a concomitant reduction in the probability of mutual gaze.

In addition, the current study aimed to provide an indirect examination of the hyper-arousal hypothesis by examining whether: 1) systematic manipulations made to the level of eye contact during instructional demands influenced the occurrence of behaviours hypothesized to be related to states of hyperarousal; 2) arousal-related behaviours were related to gaze avoidant behaviours; 3) embedding eye contact within praise served to reduce the occurrence of gaze avoidance behaviours.

The current study has a number of methodological refinements in comparison to the Murphy et al (2007) study. First, all instructional demands were presented by the experimenter, rather than via a computer, improving the ecological validity of the study. Second, regular prompts for eye contact were made in eye contact conditions, to ensure that eye contact (i.e., mutual gaze) was established, thereby improving the internal validity of the study. Third, the explicit definition of gaze avoidance conflated body/head turning and eye covering. It is possible that these behaviours serve differing functions and as such the current study aimed to examine a wider range of dependent variables. Fourth, single case experimental design methodology was adopted as the experimental paradigm. This methodology allows for examination of changes in behaviour over time at the level of the individual and allows for a more fine-grained analysis than is possible using other methodologies. Finally, the current study included both the analysis of between- and within-session data which allowed for a wider range of empirical questions to be addressed.

Methodology[20]

Participants

Four participants were recruited via the Fragile X Syndrome Society mailing list and were located within a 50-mile radius of the study centre. To meet inclusion criteria all participants were aged 5-15years old, reported by teachers or parents to have significant problems in initiating and maintaining eye contact, and to have intellectual disabilities. All participants had a confirmed diagnosis of fragile X syndrome. The first four participants who returned consent forms and met the above criteria were enrolled onto the study. All parents were visited at home to complete pre-study questionnaires. An additional school visit was also conducted for one participant (Nick) whose analysis was conducted at school.

Terry was 12 years old and communicated verbally using short sentences and engaged in stereotypical behaviours including body rocking and head rolling. Martin was 6 years old and was non-verbal and communicated primarily by gesture and engaged in stereotypical behaviours including complex finger movements and spinning objects. David was 5 years old and communicated verbally using single words and short phrases and engaged in self-injurious behaviours including banging his body against objects as well as pica, as well as aggression and property destruction. Nick was 6 years old and communicated verbally using simple phrases and words, engaged in self injurious behaviours (e.g., head hitting), stereotypy (e.g., sucking index finger), and aggressive and destructive behaviours (e.g., biting, throwing items). Table 1 shows summary information for each of the participants.

[20] The study received ethical approval from the University Ethics Committee

Table 10. *Results of Vineland Adaptive Behavior Scales, Social Communication Questionnaire and Behavior Problems Inventory*.

Measure[21]	**Terry**	**Martin**	**David**	**Nick**
Vineland Adaptive Behavior Scale (Std Scores)				
Adaptive Behaviour Composite	52	53	70	55
Communication	54	49	69	54
Daily Living Skills	57	58	73	58
Socialisation	45	55	90	65
Motor Skills	67	59	61	54
Social Communication Questionnaire (Total Score)				
	15	25	0	18
Behavior Problems Inventory Frequency (severity)				
Self-injurious behaviour	0 (0)	3 (2)	8 (5)	4 (3)
Stereotyped behaviour	22 (16)	60 (20)	4 (3)	18 (14)
Aggressive/Destructive behaviour	3 (3)	2 (1)	23 (12)	16 (16)

Setting and Materials

[21] SCQ scored out of 40. Vineland standard scores of 70 or below equate to 'low', 70-90 'moderately low', 90-110 in average range; BPI total scores for SIB frequency = 60, SIB severity = 45; Stereotypy frequency =100, Stereotypy severity = 75; Aggression frequency =48, aggression severity= 36

Pre-assessment questionnaires (see below) were completed with each participant's mother during a home visit lasting approximately 1.5 hours. The experimental analysis for Terry, Martin and David was completed in a specially designed observational room at the Cerebra Centre for Neurodevelopmental Disorders at the University of Birmingham. The room was furnished with a table and two chairs and had two cameras and audio equipment operated by a research assistant behind a one-way mirror. Preferred toys were available for each child to interact with between experimental sessions. The experimental analysis for Nick was completed in a room at his school. The room contained tables and chairs and toys and magazines were made available for Nick to interact with between experimental sessions. Each session was videotaped by an observer present in the room using a hand-held video camera.

All video footage was saved onto a removable hard-disk and coded using *Obswin* observational software (Martin, Oliver, & Hall, 2000) on a desktop computer.

Measures

The following measures (See Table 1) were completed with each participant's mother as part of the pre-study assessment; *Vineland Adaptive Behavior Scale* (Sparrow, Balla, & Cicchetti, 1984), *Social Communication Questionnaire* (Rutter, Bailey, & Lord, 2003) and the *Behavior Problems Inventory* (Rojahn, Matson, Lott, Esbensen, & Smalls, 2001).

Dependent Variables

Dependent variables were coded for both experimenter and participant behaviour using *Obswin*. All variables were recorded continuously.

The following codes were used for experimenter behaviour:

1) *Adult eye contact (duration).* Experimenter directing their gaze towards the face or eyes of participant.
2) *Verbal prompt for eye contact or alternative response (event).* Experimenter makes prompt for eye contact (e.g., "look at me!") or prompt for an alternative response (e.g.., "give me five!").
3) *Verbal prompt for participant to begin demand sequence (event).* Experimenter provides verbal prompt to complete task (e.g., "Can you match yellow?")

The following codes were used for participant behaviour.

Gaze-related variables.

1) *Looking at experimenter (duration).* Participant directs their gaze towards the face or eyes of the experimenter.
2) *Head or body turning (duration).* Participant moves their head or body 45° or more away from the experimenter and table top.
3) *Eye cover (duration).* Participant covers their face with their hands or object/surface.
4) *Eye rub (duration).* Participant rubs eyes with hands or other body part.

Previous research on fragile X syndrome has defined these four participant variables as 'gaze avoidance behaviours'. Hall, DeBernardis, and Reiss (2006), for example, provided data to suggest that eye rubbing tended to co-occur with turning and covering (defined as face hiding in that study), suggesting that these behaviours formed a response class for individuals with fragile X syndrome. Hall et al also provided evidence to suggest that these behaviours occurred at an elevated rate in an Interview condition, which included prompts for eye contact. Murphy,

Abbeduto, Schroeder, and Serlin, (2007) in their study on fragile X syndrome defined both turning and eye covering as 'gaze avoidance' behaviours.

Non-verbal signs of anxiety.

5) *Fidgeting (duration).* The participant displays restless, repetitive, non-rhythmic, non-functional motor movements, such as, moving their hands, hand wringing, touching their face or hair or moving an object, or wriggling in their seat. This code *does not* include stereotyped behaviours, which are *rhythmic*, unusual seemingly purposeless movements of their body or objects. Fidgeting has been used as an indicator of social anxiety in previous research on fragile X syndrome (Lesniak-Karpiak, Mazzocco, & Ross, 2003).
6) *Individual signs of anxiety based on parental report (duration).* Parents were asked to indicate specific behavioural topographies that they believed indicated their child was in a state of anxiety.
7) *Yawn (duration).* Participant opens mouth beyond typical opening accompanied by audible intake of breath.

It has been suggested that yawning serves the function of increasing arousal and alertness and wakefulness (Matikainen & Elo, 2008; Walusinski, 2006)[22]. Due to its hypothesized links to arousal, yawning was included as a target variable.

[22] Matikainen and Elo (2008) propose that lowered rates of yawning may indicate a higher level of baseline arousal in response to the demands of the environment, conversely higher rates of yawning may reflect a necessity to increase arousal due to the low levels of arousal in the response to the current environmental demands.

Inter-observer agreement

Inter-rater reliability was calculated for 16.66%, 20%, 30%, and 42.9% of sessions for Terry, Martin, David and Nick respectively. Kappa scores for all reported independent and dependent variables are reported in Table 2 below.

Table 11. *Kappa values for Reported Independent and Dependent Variables.*

	Terry	Martin	David	Nick
Independent variables				
Eye contact prompt	0.85	0.71	0.65	1.00
Adult eye contact	0.85	0.82	0.91	0.73
Onset of demands	0.59	0.90	0.77	0.87
Gaze Variables				
Child looking	0.81	0.73	0.65	0.72
Cover	0.48	N/A	0.87	0.66
Rub	0.65	0.40	0.84	1.00
Turn	0.58	0.41	0.61	0.51
Arousal-related				
Anxiety	0.47	0.41	0.91	0.87
Fidget	0.18	0.65	0.31	0.49
Yawn	0.79	1.00	0.87	0.72

The guidelines proposed by Fleiss (1981) for the interpretation of Kappa were adopted for the current study, with scores <0.40 regarded as indicative of poor agreement, 0.40-0.75 as indicating fair to good and scores > 75 as indicating excellent agreement. As such, all variables met criteria for fair agreement or better, with the exception of 'fidgeting' for Terry and David.

Experimental design

An alternating treatments design was used to examine the influence of eye contact during academic activities. Single case experimental design allows for the experimental analysis of the influence of an independent variable on a dependent variable over time at the level of the individual (Morgan & Morgan, 2001; Sidman, 1960) and as such was well suited to address the demands of the current research question. Two conditions (eye contact present vs. eye contact absent) were alternated in a randomized fashion. Randomized sequences were generated using a web-based randomizer programme.

Procedure

Analysis of effects of manipulating eye contact.

The participant and the experimenter were seated together at a table. Tasks were identified following discussion with the child's teacher and tasks for all participants formed part of each child's IEP and were selected because the child was likely to have some familiarity with them but struggled to complete them independently without assistance. The task selected for Terry involved completing pre-school arithmetic worksheets involving simple addition, for Martin and Nick tasks involved matching buttons of different colours and shapes and for David involved the same matching button task but also involved threading.

In each condition, demands were presented in a discrete trial format using a standard three-step prompting procedure (Horner & Keilitz, 1975). An initial verbal prompt was provided; if accurate compliance did not follow within 5s then the correct response was modelled by the experimenter; if the request was not complied with, then the child was supported to complete the task using hand-over-hand guidance. Praise for accurate performance was provided if the child completed the task following verbal or modelled prompts. If hand-over-hand prompting was required then the experimenter stated "Ok let's try the next one!". All problem behaviour was ignored[23], except for elopement, whereby the experimenter would guide the participant back to their chair using minimal interaction.

Session length varied between participants and was based on how long the child was expected to work in natural classroom settings. For Terry sessions lasted 10 minutes, for Martin and David sessions 5 minutes and for Nick approximately 3 minutes. With the exception of Nick, demands were presented following a fixed time (FT) schedule, at a rate of no greater than FT-30s (i.e., no more than 20 trials over 10 minute period) to control for the rate of demand presentation (e.g., Smith, Iwata, Goh, & Shore, 1995). The experimenter was prompted by the research assistant as to when to begin the next trial using a 'bug in the ear' device. The number of sessions completed for each participant was dependent on the number that could be completed in the allotted time available for each participant (typically across three visits lasting up to an hour each).

[23] Criteria for session termination were established with parents and teachers in advance for each participant however these criteria were never reached for any participant.

Some modifications to the protocol were required for Nick. Specifically, demands were presented without any inter-trial delay and preferred foods/items were presented as a form of reinforcement at the end of each session. Both these modifications formed a typical part of the teaching strategies used to support Nick in his typical classroom setting and were required in order to facilitate his inclusion in the current study.

Eye contact condition.

In the eye contact condition, the experimenter sat directly opposite the child. Before the session began the experimenter stated, "Remember to keep looking at me when we do your work!". To further aid discrimination between the conditions a red table top cover was laid out. Levels of eye contact were manipulated prior to, during and consequent to each discrete trial. Prior to the onset of each trial, the experimenter presented a prompt for eye contact "Ok, look at me", using a two-step prompting procedure (verbal prompt, model). The experimenter then attempted to fix his gaze on the child's eyes at the beginning of the trial and continue looking at the child as they completed the task. At the end of the trial, the experimenter directed their gaze towards the child's whilst delivering verbal praise or a prompt to move on to the next trial. The mean level of eye contact provided by the experimenter towards the participant in this condition for Terry was 91.32% (range = 80-98.37%); for Martin was 81.03% (range = 74.19-83.61%); for David was 94.2% (range = 90.48%-98.33%); for Nick was 91.61% (range =88.46-100%).

No eye contact condition.

In the no eye contact condition, the experimenter sat adjacent to the child. Prior to the session beginning the experimenter stated, "You don't need to look at me when we do your work". To further aid discrimination between conditions a white tabletop cover was laid over the

table. In order to control for the potential rate of demands, a single prompt for an alternative motor response (e.g., “touch your nose”, “give me five”, “pick up the pen”) was delivered antecedent to each trial, using the same two-step prompting procedure. During the trial, the same prompting procedures were used as in the *Eye Contact* condition with the exception that the experimenter did not direct his gaze towards the participant. After each trial, no eye contact was provided when delivering praise or a prompt to move on to the next trial. The mean level of eye contact provided by the experimenter towards the participant in this condition for Terry was 8.19% (range = 3.15-14.05%); for Martin was 2.00% (range = 0-6.76%); for David was 3.48% (range = 1.61%-7.5%); for Nick was 22.57% (range = 3.77-37.93%).

Data analysis

Data were reduced to presence vs. absence in 5s epochs for the purpose of sequential analysis and thus the data analysis was analogous to the use of a 5s partial interval recording system. To overcome difficulties with reliance on visual inspection (Wolery, Busick, Reichow, & Barton, 2010), the dominance statistic (*d*; Cliff, 1993) was calculated for each participant’s target behaviour across each experimental condition. The *d* statistic measures the extent to which one sample distribution lies above or below another and is calculated by comparing all scores in one condition to all scores in another using a dominance matrix. A value of +1 was allocated if the value of the eye contact datum point was greater than the value of the no eye contact datum point. A value of -1 was allocated if the value of the eye contact datum point was lower than that of the no eye contact datum point. A zero was allocated if the two values were identical. As such a *d* value of +1 indicates that each datum point is greater than every other datum point in another

series, whereas a *d* value of -1 indicates that each datum point in a series is less than every other datum point in another series. A *d* value of between +/- 0.33 was used to indicate differentiation.

A series of duration-based lag sequential analyses were completed to examine within-session trends in the data. These analyses were needed to clarify the nature of the relationship between variables. Yule's Q (Bakeman & Quera, 1995; Hall & Oliver, 1997) was used to analyse the conditional probability of the association between criterion and target variables (analogous to independent and dependent variables respectively). Yule's Q provides a measure of association between an event and a response, whilst controlling for chance. Yule's Q scores were calculated for the 5s lag that co-occurred with the onset of the criterion variable, as well as the three 5s lags both antecedent and consequent to its onset. Yule's Q values range from -1 to +1. A positive value indicates the conditional probability of the target variable is higher than would be expected by chance, and a negative value for Q indicates that the conditional probability of the target variable is lower than would be expected by chance. A score of +/-.33 was used to indicate significance (depicted by a 'x' on all lag sequential analysis graphs), which equates to an odds ratio of 2, indicating that a behaviour is twice as likely to occur in the presence of another behaviour or environmental condition (Bakeman & Quera, 1995; Hall & Oliver, 1997). All analyses were restricted to analyse the target variable up until the next occurrence of the criterion variable, in order to avoid a confounding effect from independent variables occurring in close proximity.

Results

The Effect of Eye Contact on Gaze Avoidance Behaviours.

Between-session data were analysed to determine the nature of the relationship between eye contact and forms of gaze-avoidance behaviours. Appendix 1 presents between-session data for all topographies of gaze-avoidance behaviours. Figure 2 presents between-session data for all topographies of gaze-avoidance behaviours that met criteria for differentiation (i.e., *d* = or > .33).

For Terry eye covering occurred at differentially higher rates in the eye contact condition (Cliff's *d* score of 0.83; Eye contact, mean = 11.23, range =3.97-15.45; No eye contact, mean = 3.9, range = 0.79-10.4). Eye covering and eye rubbing did not appear to be any more likely to occur in the eye contact condition (Cliffs *d* <0.33). For Martin, eye rubbing occurred at differentially higher rates in the eye contact condition (Cliff's *d* score of 0.92; Eye contact, mean 17.73, range =9.68-23.33; No eye contact, mean = 7.11, range = 0-10.17), no such relationship was revealed for turning (Cliff's *d* score < 0.33). For David, turning occurred at differentially higher rates in the eye contact condition (Cliff's *d* score of 1.0; Eye contact, mean = 41.31, range = 33.33-54.1; No eye contact, mean = 10.89, range = 6.45-15). Between-session data did not reveal any differentiation for eye covering (Cliff's *d* score of <0.33). For Nick, between-session data did not reveal any differentiation between eye contact and no eye contact conditions for eye rubbing and eye covering. Data for turning showed considerable overlap between experimental conditions, but met criteria for differentiation (Cliff's *d* score of 0.33; Eye contact, mean = 31.47, range = 13.64-50; No eye contact, mean = 17.78, range = 5.66-34.48).

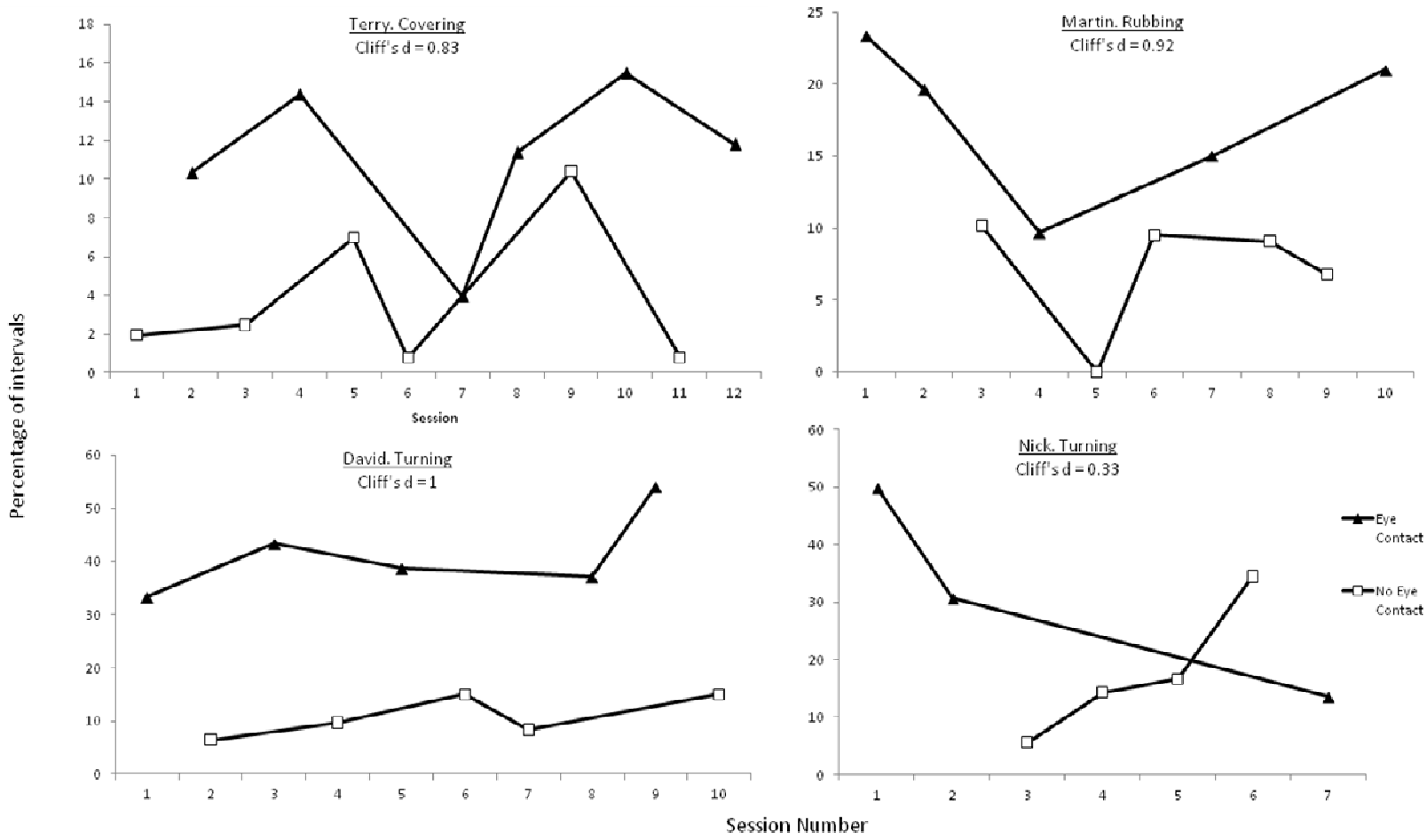

Figure 2. Differentiated topographies of gaze-avoidance behaviour in eye contact and no eye contact conditions (see Appendix 1 for full data set).

Co-occurrence of Different Topographies of Gaze-Avoidance Behaviours

Lag sequential analysis, depicted in Figure 3, indicated a relationship between different topographies of gaze-avoidance behaviours for both Terry and David. For Terry there appeared to be an increase in the conditional probability of eye rubbing following the onset of eye covering. No such relationship was found between turning and eye covering. As such eye covering and eye rubbing were combined to form the variable 'target gaze-avoidance behaviours' for subsequent analyses for Terry. For David, there appeared to be an increase in the conditional probability of turning following the onset of eye covering. No such relationship was identified between turning and eye rubbing. As such, these two variables were combined for subsequent within-session analyses to form the variable 'target gaze-avoidance behaviours' for David[24].

[24] No such relationships were identified for either Martin or Nick. As such, eye rubbing was selected as the target gaze-related behaviour for all subsequent within-session analyses for Martin and turning for Nick for all subsequent analyses.

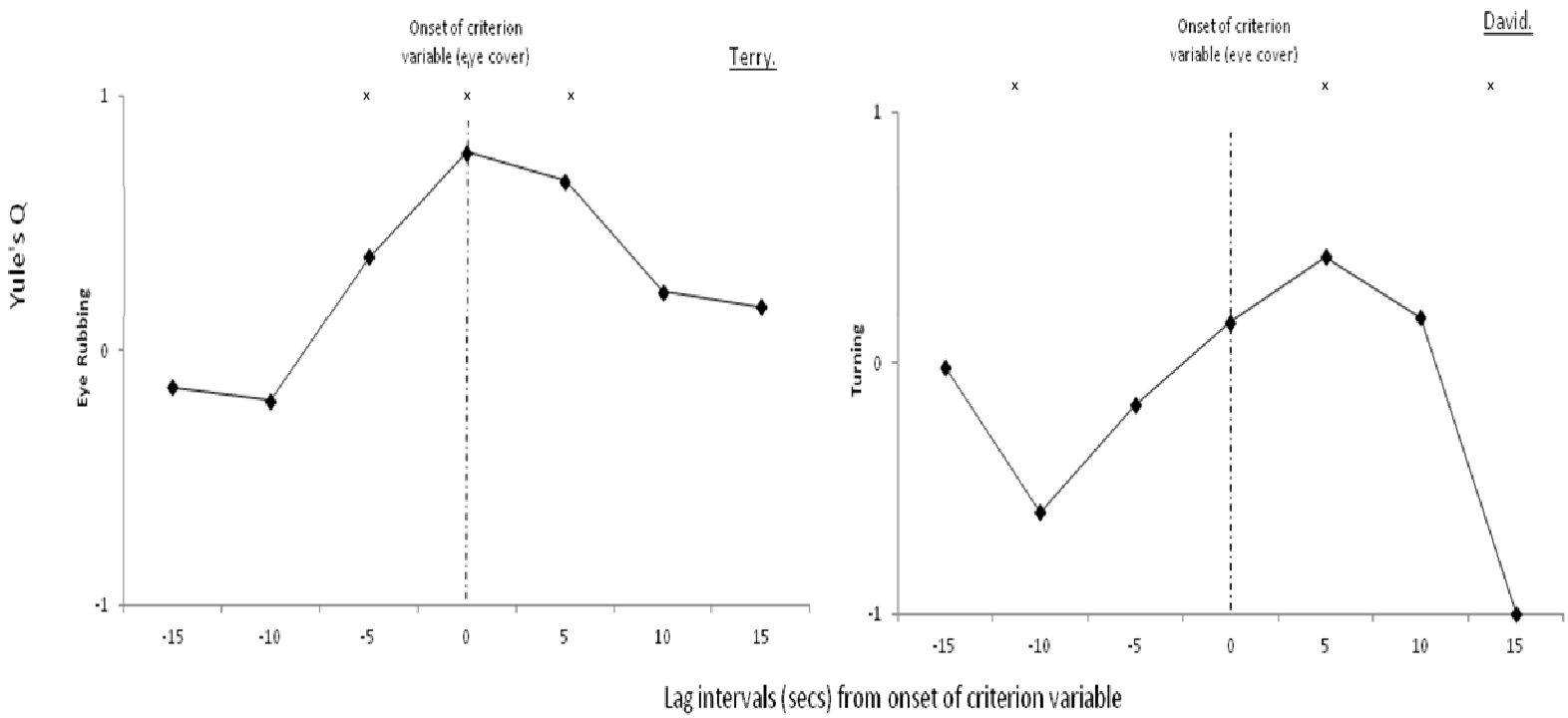

Figure 3. Lag sequential analysis depicting relationship between different gaze-avoidance behaviours for Terry and David.

Aim III. Are Target Gaze-Avoidance Behaviours Evoked by Prompts for Eye Contact?

A series of lag sequential analyses were run to determine whether the conditional probability of target gaze avoidance behaviours was raised following the onset of prompts for eye contact. Figure 4 depicts the results of these analyses for each participant.

For Terry and Martin, the conditional probability of target gaze avoidant behaviours appears to rise following the onset of a prompt for eye contact. For Nick there appears to be an increased probability of such behaviours occurring within the same epoch as the onset of the prompt for eye contact. This pattern of results indicates that such behaviours may be evoked by prompts for eye contact.

The lag sequential analysis for David shows that the probability of target gaze avoidance behaviours reduces following the onset of prompts for eye contact, which appears somewhat paradoxical. The raised conditional probability of these behaviours in the three lags preceding the onset of the prompt for eye contact may indicate that these behaviours served an avoidance, rather than escape function.

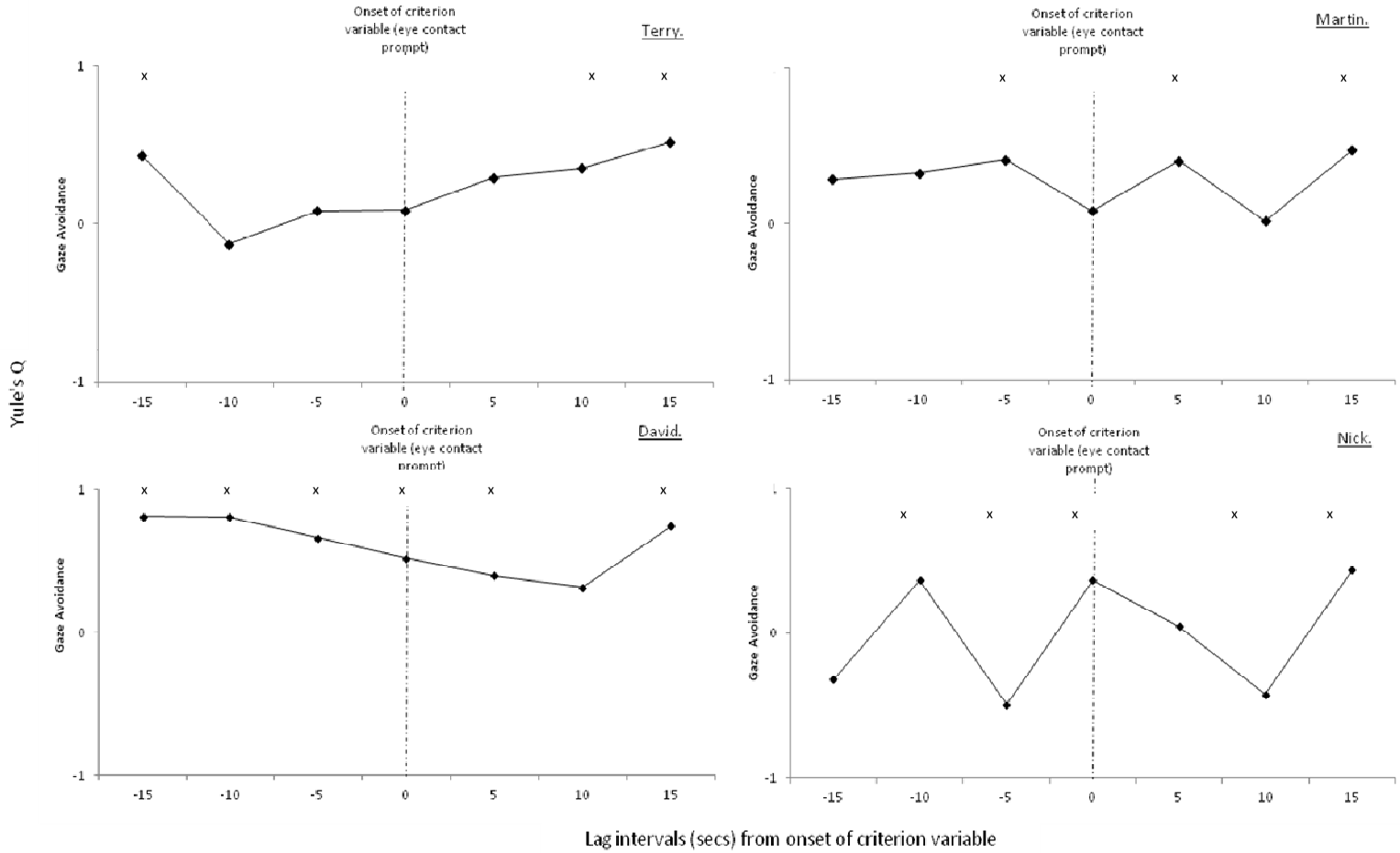

Figure 4. Lag sequential analysis depicting conditional probability of gaze avoidance given onset of eye contact prompts.

Examination of the Function of 'Target Gaze Avoidance Behaviours'

Figure 5 depicts the results of the lag sequential analysis which examined whether the onset of task-related demands was associated with an increase in the conditional probability of gaze-avoidance behaviours.

The lag sequential analysis for Terry and Martin do not provide any evidence to suggest a relationship between target gaze avoidance behaviours and the onset of task-related demands. Indeed, for both participants the conditional probability of gaze avoidance appears to reduce at Lags 0 and 5s. The lag sequential analysis for David shows a similar pattern of results for the onset of prompts for eye contact, with a heightened probability of target behaviours occurring in the three lags preceding the onset of the task-related demand sequence. It is unclear, for David, therefore whether target gaze avoidance behaviours occur to avoid prompts for eye contact or to avoid the onset of the demand sequence. For Nick there does not appear to be an increase in gaze avoidance behaviours following the onset of the task-related demand sequence.

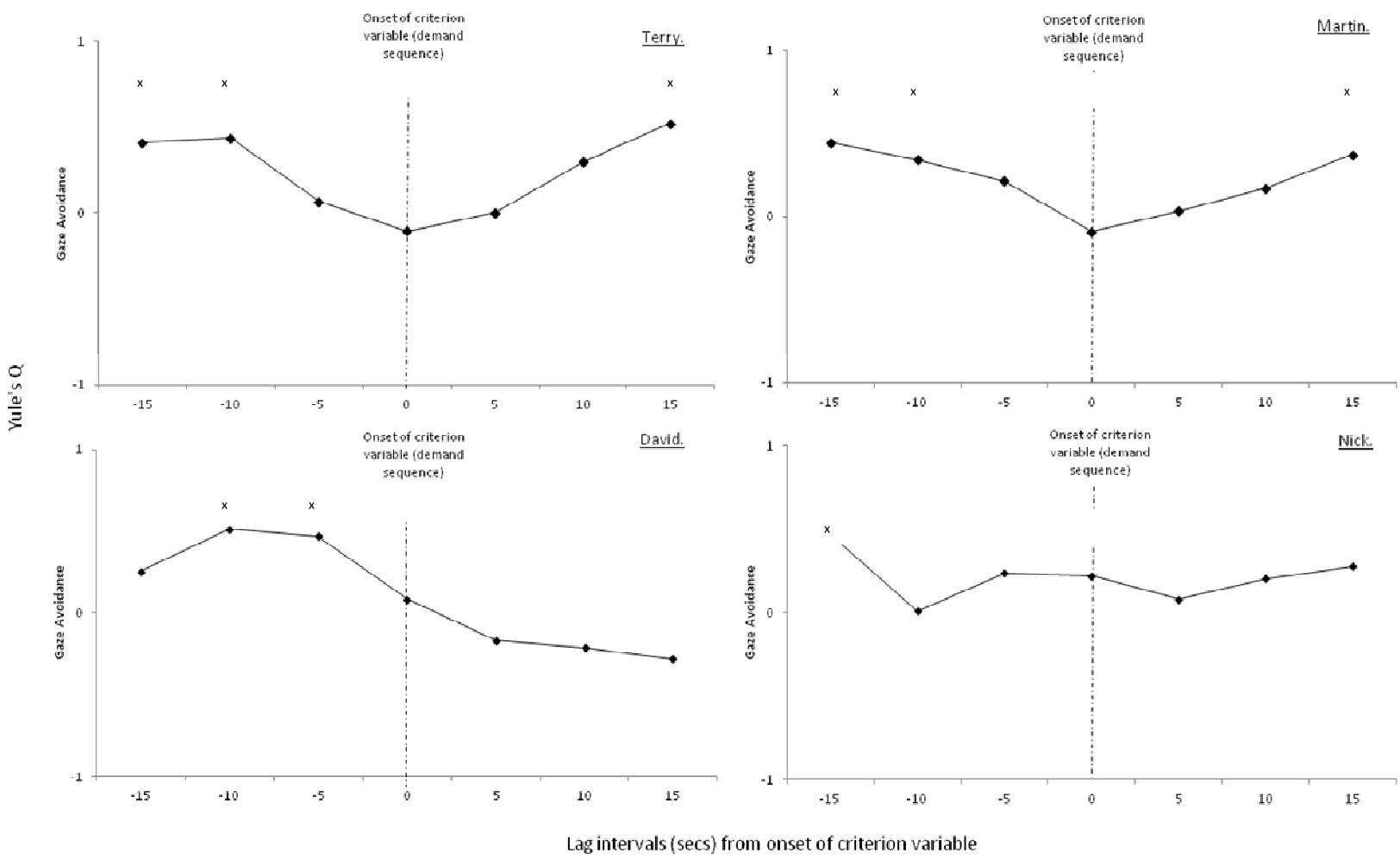

Figure 5. Lag sequential analysis depicting conditional probability of gaze avoidance given onset of demand sequence.

Aim V. Is the Probability of Mutual Gaze Reduced Following the Onset of Target Gaze Avoidance Behaviours?

The variable 'mutual gaze' was created by combining the variables Adult Eye Contact and Participant Eye Contact for all instances of simultaneous occurrences. A series of lag sequential analyses, depicted in Figure 6, were conducted to determine whether the occurrence of target gaze avoidance behaviours were temporally contiguous with a reduction in the probability of mutual gaze. It was hypothesised that this would be indicative of a putative negative reinforcement contingency if the behaviours served the function of escaping from eye contact.

There appears to be a reduction in the conditional probability of mutual gaze following the onset of target gaze avoidance behaviours for Terry, Martin and Nick respectively. This suggests the presence of a potential source of negative reinforcement (i.e., removal or attenuation of eye contact) that is temporally contiguous with the onset of target gaze avoidance behaviours. For David, however, there does not appear to be a reduction in the conditional probability of mutual gaze following the onset of target gaze avoidance behaviours. It may be that these behaviours are therefore related to task-related demands rather than the prompts for eye contact per se.

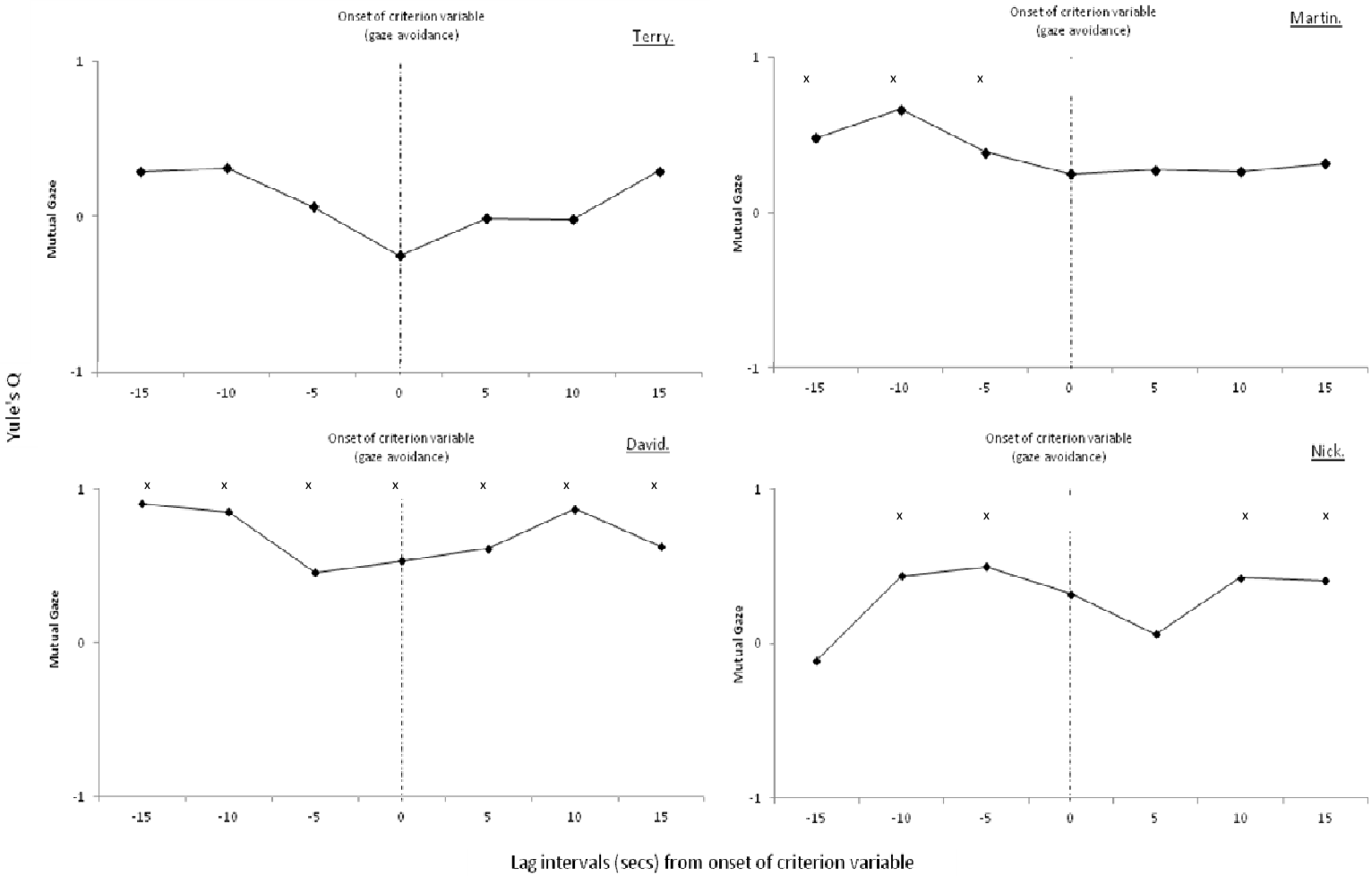

Figure 6. Lag sequential analysis depicting conditional probability of mutual gaze given onset of gaze avoidance behaviours.

Relationship Between Eye Contact and Arousal-Related Behaviours?

Between and within-session data were examined to determine the nature of the relationship between eye contact and putative signs of arousal. Appendix 2 shows the results of the between-session data for these analyses.

As shown in Appendix 2 there was considerable inter and intra-individual variability in the pattern of results found for arousal-related variables (reported indicators of anxiety, fidgeting and yawning). Figure 7 shows between-session data for topographies of fidgeting and indices of anxiety that met criteria for differentiation (*d*= or > 0.33). Terry showed slightly elevated rates of fidgeting in eye contact conditions, which met criteria for differentiation (Cliff's *d* = 0.33; Eye contact, mean = 50.24, range = 38.62-72; No eye contact, mean = 41.02, range = 24.03-57.02). There did not appear to be any difference between conditions for reported signs of anxiety. Martin, David and Nick all showed elevated rates of signs of anxiety under eye contact conditions. Indeed Martin had a Cliff's *d* of 0.60 (vocalisations; Eye contact, mean = 32.38, range = 3.23-62.9; No eye contact, mean = 10.74, range = 1.52-34.33)[25], David of 0.68 (thumb sucking; Eye contact, mean = 42.11, range = 0-85.71; No eye contact, mean = 5.98, range = 0-20) and Nick of 0.58 (fingers in mouth; Eye contact, mean = 19.44, range = 0-33.33; No eye contact, mean = 0.94, range = 0-3.77).

[25] Somewhat paradoxically and as shown in Appendix 2, Martin displayed elevated rates of fidgeting in No Eye Contact conditions, with a Cliff's *d* of -0.52 (Eye Contact, mean = 11.58, range = 3.28-30; No Eye Contact, mean = 19.50, range = 10.17-28.57).

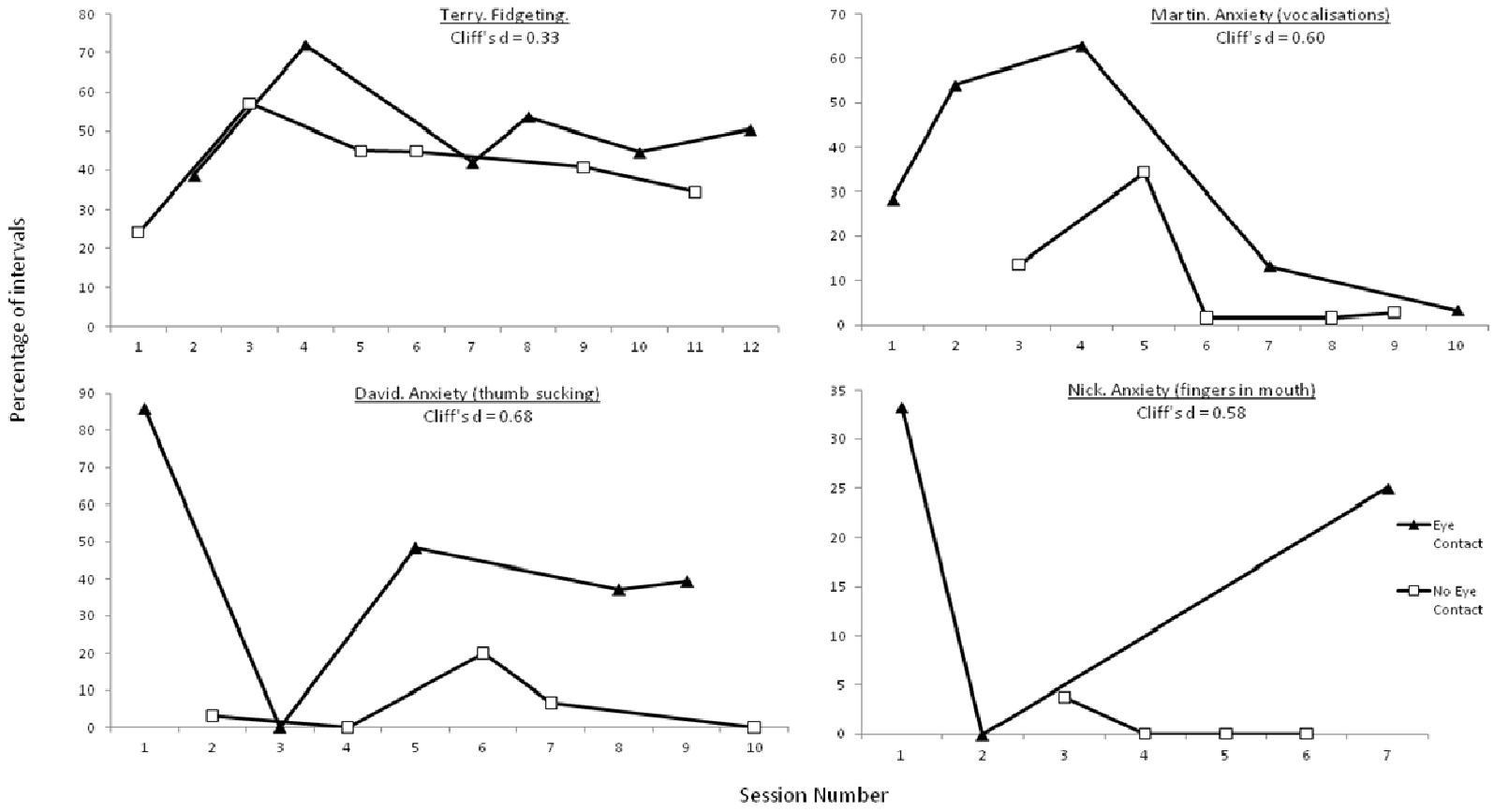

Figure 7. Differentiated topographies of arousal-related behaviour in eye contact and no eye contact conditions (see Appendix 2 for full data set).

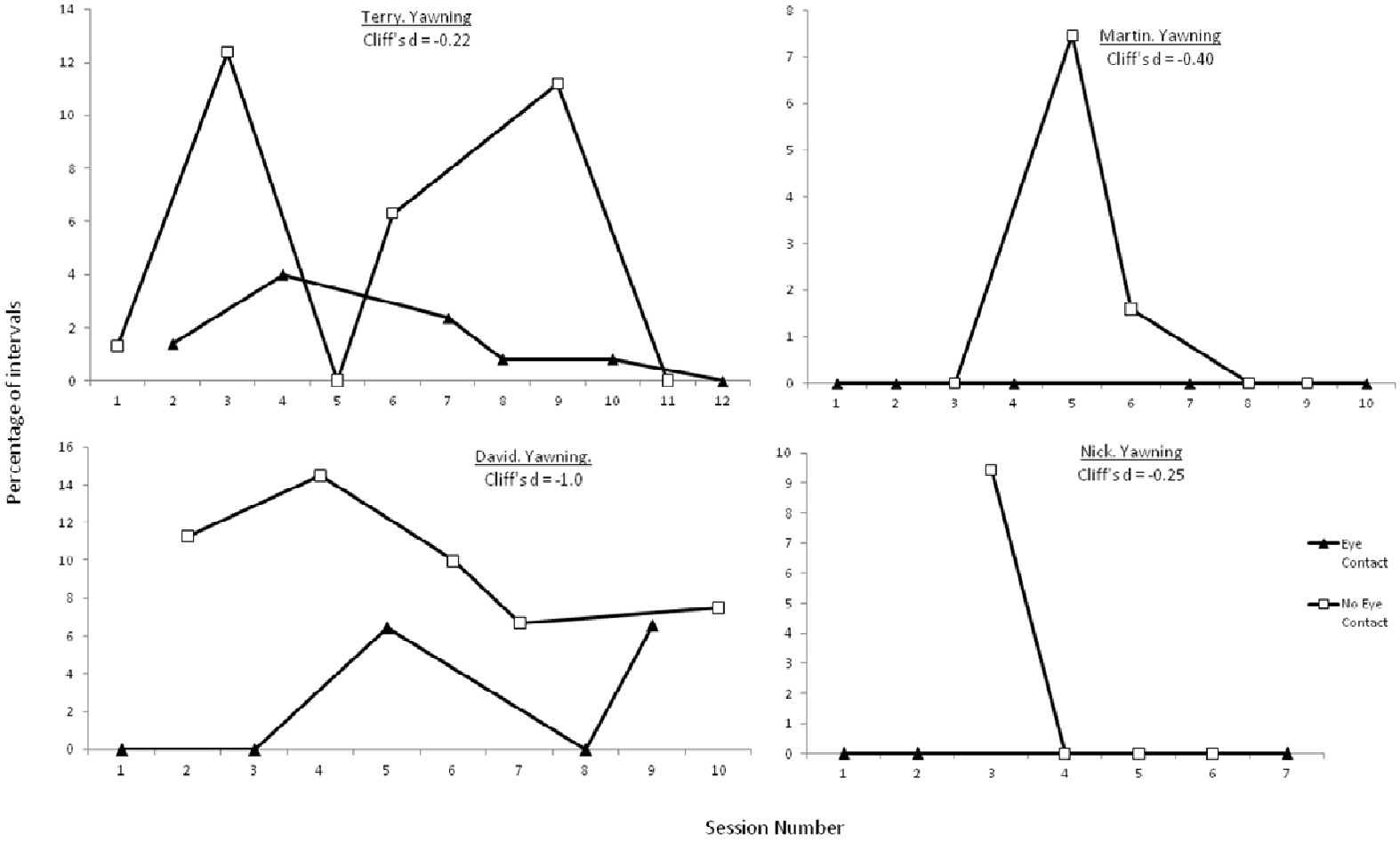

Figure 8. Between-session occurrence of yawning in eye contact and no eye contact conditions.

Figure 8, shows between-session data for yawning for all participants. Yawning appeared to occur at reduced levels in Eye Contact conditions for all participants. For Terry there were lower rates of yawning in the Eye Contact condition (Cliff's *d* = -0.22; Eye Contact, mean = 1.56, range = 0-4; No Eye Contact, mean = 5.2, range = 0-12.4). Similar results were reported for Martin (Cliff's *d* = -0.40; Eye Contact, mean =0; No Eye Contact, mean = 1.81, range = 0-7.46); David (Cliff's *d* = -1.0; Eye Contact, mean = 2.6, range = 0-6.56; No Eye Contact, mean = 9.99, range = 6.67-14.52); and Nick (Cliff's *d* = -0.25; Eye Contact, mean =0; No Eye Contact, mean = 2.36, range = 0-9.43). This pattern of results complements the data presented for anxiety and fidgeting as it is hypothesised that yawning reflects a lower state of arousal and as such may be incompatible with an anxiety related response.

Aim VII. Nature of the Relationship between Gaze Avoidance Behaviours and Indices of Hyper-arousal (Fidgeting/Reported Signs of Anxiety).

Figures 9 and 10 depict the results of the lag sequential analysis for fidgeting and reported indices of anxiety respectively.

Figure 9 suggests that for Terry, David and Nick the onset of fidgeting co-occurs with an elevation in the conditional probability of target gaze avoidance behaviours at lag 0 (relative to lags – and + 5s), indicating a relationship with the onset of fidgeting. These data appear to suggest that these target gaze avoidance behaviours are associated with the onset of an index of anxiety. The data for Martin suggest an inverse relationship between target gaze avoidance behaviours and fidgeting.

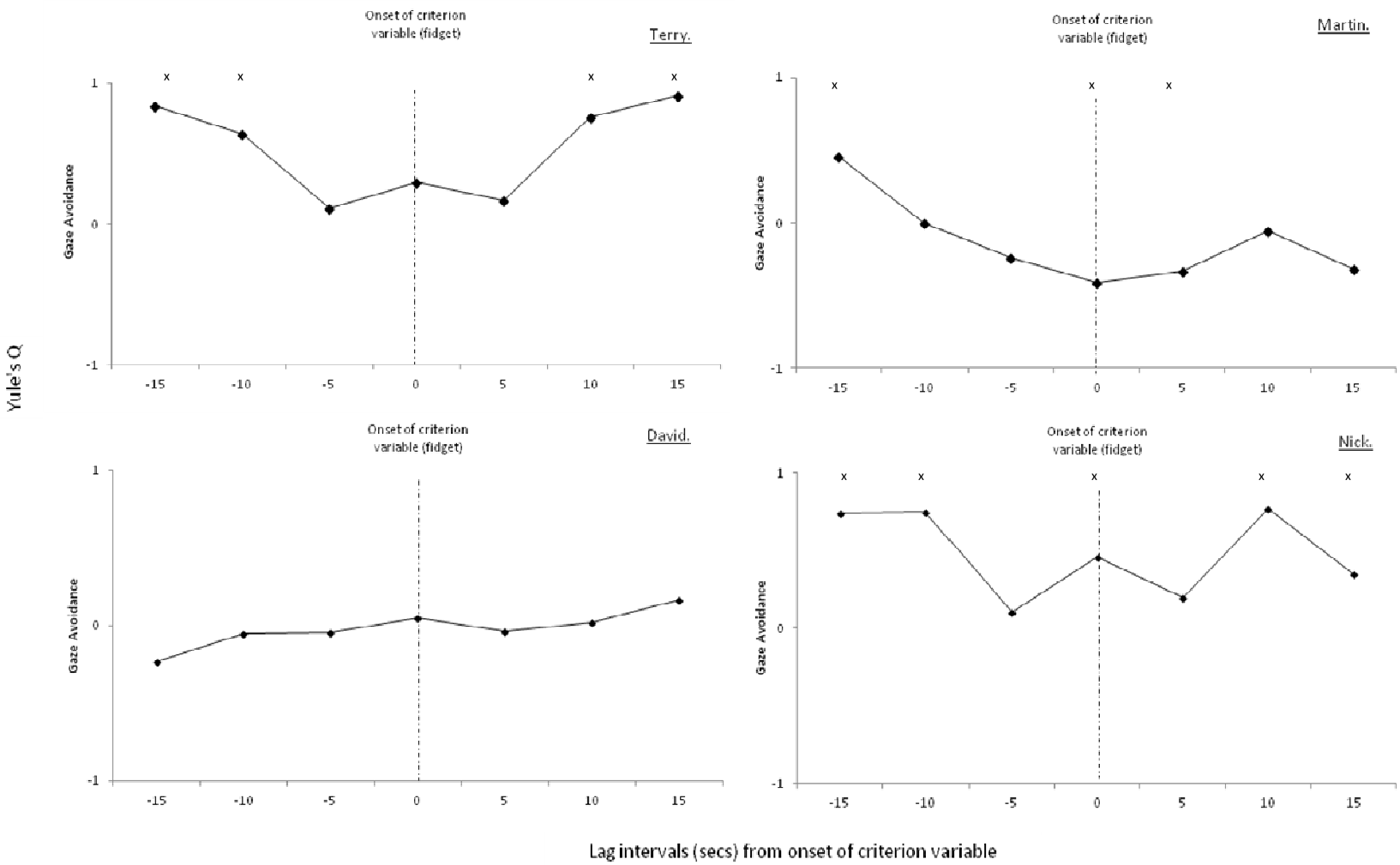

Figure 9. Lag sequential analysis depicting conditional probability of gaze avoidance behaviours given onset of fidgeting.

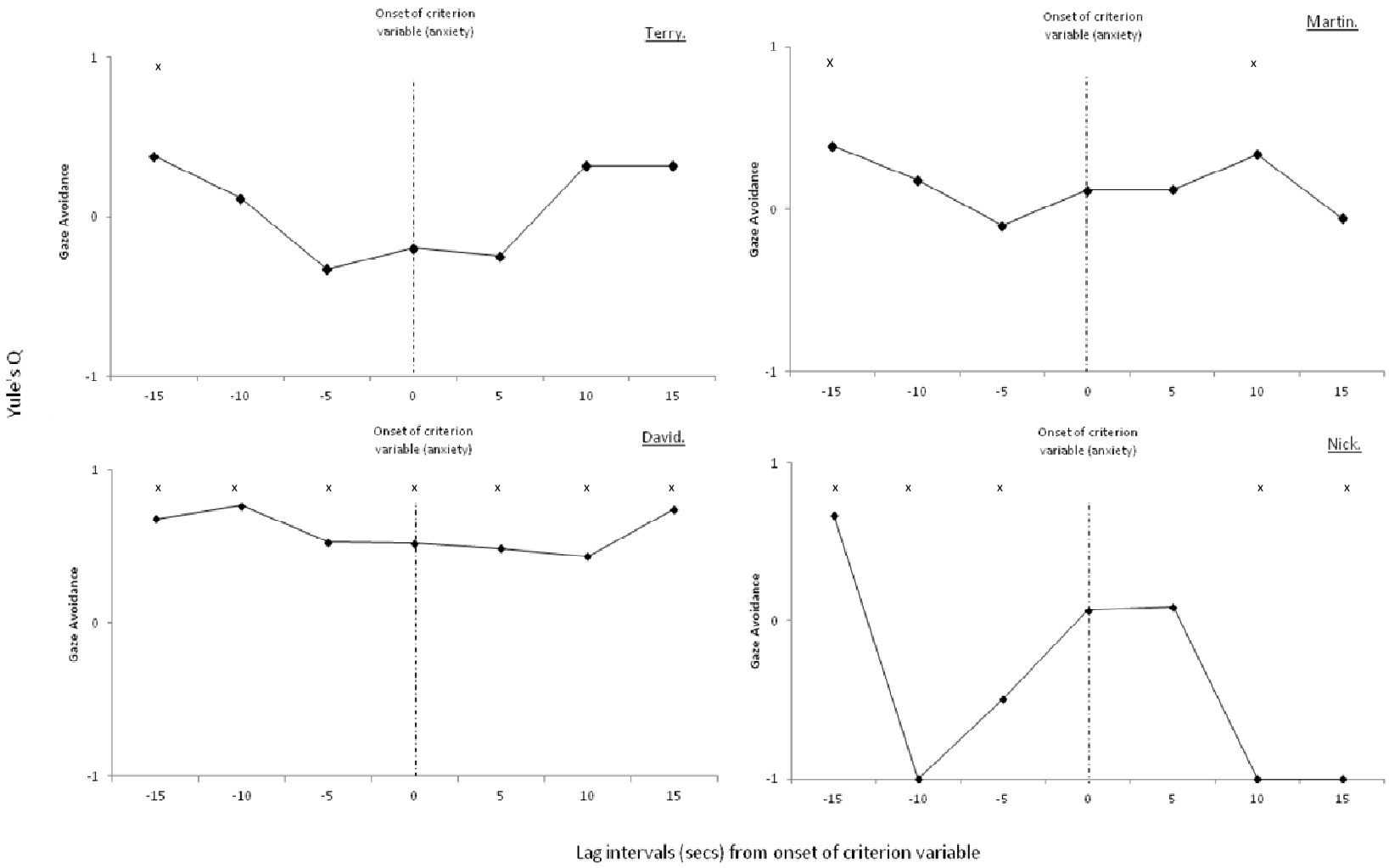

Figure 10. Lag sequential analysis depicting conditional probability of gaze avoidance behaviours given onset of reported indices of anxiety.

The data from the lag sequential analysis for Martin, depicted in Figure 10, suggest an increase in target gaze avoidance behaviours following the onset of the 'anxiety' related response. Data for Terry suggests an inverse relationship between target gaze avoidance behaviours and the onset of the reported indices of anxiety. Data for David do not indicate any relationship between these two variables, perhaps reflecting the enduring nature of the target behaviour (thumb sucking and fingers in mouth).

Aim VII. Are There Conditions That Reduce the Probability of Target Gaze Avoidance Behaviours?

It was deemed of interest to examine whether target gaze avoidance behaviours were equally likely to occur when eye contact was paired with the delivery of praise. A reduction in the probability of gaze avoidance behaviours under such conditions would suggest that praise serves to abolish the aversiveness of mutual gaze.

As depicted in Figures 11, the lag sequential analyses for Terry, Nick and David suggest that when eye contact was paired with praise there was no increase in the probability of target gaze avoidance behaviours. Indeed, for Terry and David there appears to be a marked reduction in the conditional probability of these behaviours at lag 0. The lag sequential analysis for Martin shows a different pattern of results, suggesting that praise was associated with an increased probability of target gaze avoidance behaviours at lag 0 relative to lag – 5s..

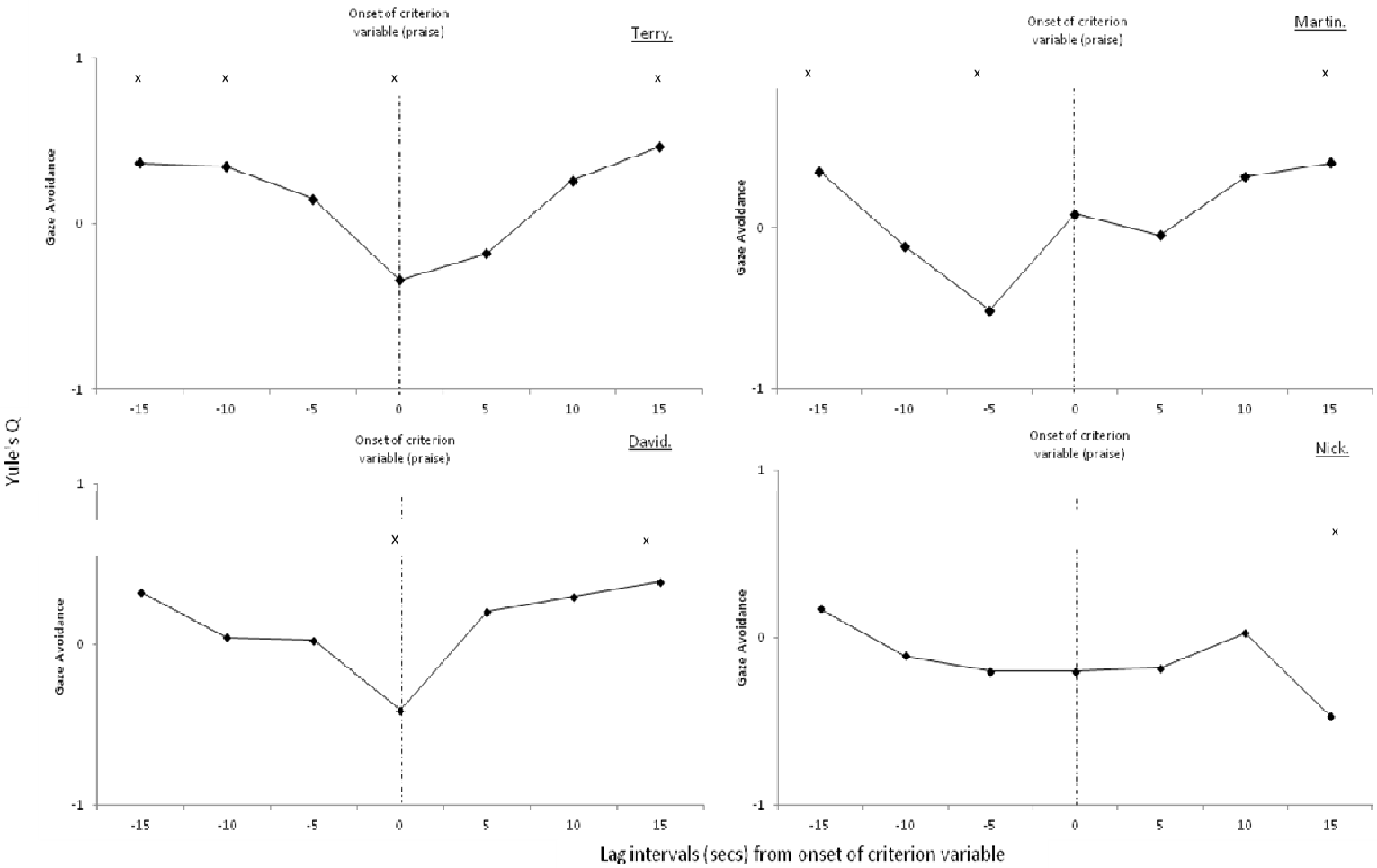

Figure 11. Lag sequential analysis depicting conditional probability of gaze avoidance behaviours given onset of praise.

Discussion

The current study examined the function served by gaze avoidance behaviours in FXS under controlled experimental conditions. All participants presented with specific topographies of gaze avoidance behaviours that were differentially elevated under high eye contact conditions. For two participants these appeared to form part of a response class hierarchy with other topographies of gaze avoidance behaviours. Data for three participants (Terry, Martin and Nick) suggested that the elevated rates of target gaze avoidant behaviours that occurred in the eye contact condition may have, at least in part, been maintained by the attenuation or removal of eye contact. No such relationship was found for David, suggesting that the elevated rates of turning that occurred in the Eye Contact condition was not related to the removal of eye contact *per se*.

A secondary aim was to examine the relationship between gaze avoidance behaviours and indices of arousal/anxiety. Despite there being considerable within- and between-subject variability, the between-session data indicated that all four participants showed differentially elevated rates of *either* fidgeting *or* reported signs of anxiety in the eye contact condition. Lag sequential analyses showed evidence of an increased probability of target gaze avoidance behaviours, following the onset of fidgeting for Terry, David and Nick and of reported indices of anxiety for Martin. Finally, there were lower rates of yawning across eye contact conditions for all participants, which suggests that the level of arousal in the eye contact condition may have been higher than in the no eye contact condition. These findings would seem to suggest that: 1) levels of arousal are elevated in high eye contact conditions and 2) markers of arousal are related to the occurrence of gaze avoidance behaviours. For two participants (Terry and David) pairing eye contact with praise appeared to have an abative impact on target gaze avoidance behaviours.

This suggests that the context in which eye contact is provided in FXS plays a critical part in establishing or abolishing its aversiveness.

These findings contribute to the existing literature on gaze avoidance behaviours in FXS. Historically there has been an implicit assumption that topographies of behaviour associated with FXS, such as turning, eye rubbing and eye covering, are maintained by the avoidance of eye contact (Wolff, Gardner, Paccia, & Lappen, 1989). That is, the function of the behaviour has been inferred from the topography. As noted by Hall, Maynes and Reiss (2009) however, without manipulating the contingencies available for gaze aversion during a functional analysis, it is difficult to provide a 'believable demonstration' of the function served by such behaviours. Competing hypotheses regarding the function of such behaviours have been postulated. Murphy et al (2007) for example, after finding little differentiation in gaze avoidance behaviours between a condition in which a task was presented by an experimenter and a computer, have suggested that it is the sensory and/or cognitive demands associated with eye contact that serve to evoke gaze avoidance behaviours in FXS, rather than eye contact itself. In partial support of these findings, Hall, DeBernadis and Reiss (2006), reported higher rates of face-hiding (including turning and eye covering) in conditions in which males with FXS were asked to sing, than an interview condition which included frequent prompts for eye contact[26]. The findings from the current study, suggest that some topographies of behaviour previously hypothesised as constituting 'gaze avoidance', show little differentiation between conditions in which eye contact is present or absent. This suggests that for some individuals with FXS, specific topographies of 'gaze avoidance behaviours' are unrelated to the presence of eye contact and are presumably a

[26] Data for eye rubbing showed the opposite pattern of results however.

function of the task demands or some other prevailing feature of the social context. In contrast, other topographies of 'gaze avoidance behaviours' showed clear differentiation under conditions of high eye contact, suggesting that the presence of eye contact was directly related to the occurrence of these behaviours. Within-session analyses suggested that these specific topographies occurred *either* as a means of attenuating mutual gaze following the onset of a prompt for eye contact *or*, as for David and potentially Nick, the presence of eye contact served to increase the aversive nature of the task and evoke 'gaze avoidance' behaviours.

Aversive stimuli typically activate the sympathetic nervous system (SNS). The hyper-arousal of the SNS is a core feature of the behavioural phenotype of FXS. For example, Miller et al (1999) reported greater electrodermal response and lower habituation to a range of sensory stimuli in FXS in comparison to age and gender-matched controls. Hall, Lightbody, Huffman, Lazzeroni, and Reiss (2009) have reported that, in comparison to siblings, males with FXS show higher heart rate, lower amplitude respiratory sinus arrhythmia (RSA) and lower heart-rate variability both at baseline but also during a social interaction. This pattern of results indicates the over-activation of the sympathetic nervous system and lower activation of the parasympathetic nervous system in participants with FXS. However, the association between this and gaze avoidance behaviours in FXS is unclear. In an uncontrolled study, Belser and Sudhalter (1995) reported an association between higher levels of skin conductance response (an indicator of sympathetic nervous system arousal) and conditions that involved high levels of eye contact. In their study, however, Hall, Lightbody, Huffman, Lazzeroni, and Reiss (2009) reported that gaze avoidance behaviours reduced over the course of a 25 minute intensive social interaction with an unfamiliar experimenter (i.e., participants evidenced the 'warm up' effect) and did not

appear to be related to cardiovascular activity. The findings of the current study suggested an association between specific behavioural indices of anxiety/arousal and gaze avoidance behaviours, although the nature of these varied substantially between participants. In addition, all participants showed elevated levels of yawning in the No Eye Contact condition, which appears to complement these findings. Hypotheses regarding the function of yawning (Matikainen & Elo, 2008; Walusinski, 2006), suggest that it occurs in contexts in which levels of arousal need to be increased in order to meet the demands placed by the environment (i.e., being repeatedly asked to complete academic tasks). If levels of arousal are already raised (i.e., by the repeated prompts for eye contact) then yawning is not required in order to increase arousal levels. The current study therefore would seem to provide some tentative evidence to support an association between: 1) higher levels of eye contact and indices of arousal and 2) indices of arousal and gaze avoidance behaviours.

In the wider literature, the social significance of direct eye contact has attracted increased attention, this literature may have implications for the understanding of gaze avoidance in FXS. The amygdala is thought to play an important role in evaluating the salience of perceived gaze direction and in mediating the affective arousal elicited by eye contact (Senju & Johnson, 2009). It has been argued that the affective arousal elicited by eye contact influences subsequent perceptual and cognitive processing (Senju & Johnson, 2009). Helminen, Kaasinen, and Hietanen (2010) demonstrated that direct gaze evoked higher skin conductance responses than averted gaze or closed eye conditions independent of the duration of eye contact. Therefore, evidence exists to support the hypothesis that even brief eye contact increases ANS arousal in the general population. Studies have reported on the association between social anxiety and the perception of

direct eye contact as threatening (Schneier, Rodebaugh, Blanco, Lewin, & Liebowitz, 2011; Wieser, Pauli, Alpers, & Muhlberger, 2009). Skuse (2003) has argued that due to its evolutionary history as a signal of threat, eye contact automatically elicits an amygdala-dependent fear response, mediated by the 'phylogenetically ancient' subcortical neural system. This fear response is, in adult humans, subsequently controlled by the 'phylogentically recent' neocortical inhibitory pathways[27].

These findings and theoretical perspectives raise interesting possibilities for the understanding of gaze aversion in FXS. That is: 1) due to its evolutionary history as a threat-related stimulus, direct eye contact elicits an amgydala-related response in humans. The amygdala is associated with the emotion of fear and displays of defensive behaviour (e.g., avoidance, aggression) and plays a central role in fear conditioning (LeDoux, 2000), 2) when humans perceive the presence of a threat direct eye contact is more likely to be perceived as threatening and presumably gaze aversion is more likely to occur (Wieser, Pauli, Alpers, & Muhlberger, 2009), 3) more recently, in evolutionary terms, eye contact has also evolved to play an important role in mediating human social approach behaviours and in the establishment and

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"One critical 'threat stimulus' is direct eye contact, but this is handled in a unique way by the human neocortex, for we have harnessed the arousal engendered by such contact for a variety of purposes that are critical to the survival of our species, including the attachment between parent and infant and pair-bonding between adults. In order to achieve this relatively recent (in evolutionary terms) adaptation of a phylogenetically ancient neural system, we have developed systems of reciprocal control over amygdala activity – exercised in particular by frontocortical circuits involving the occipito-frontal cortex, the anterior cingulate cortex, and the insular cortex. Uniquely among all other species, we are able to control this complex 'survival system' - and we do so by means of a variety of inhibitory pathways, linked into memory and language centres of bewildering complexity. Possibly because of their relatively recent evolutionary origins, the inhibitory systems are liable to dysfunction – and when they are dysfunctional one possible outcome is an impact on quintessentially human traits of social cognition." (Skuse, 2003, p. 57).

maintenance of relational and social bonds with others. As such, phylogenetically newer brain systems have developed to facilitate social approach in response to direct eye contact and to inhibit the 'threat-related' response of the amygdala and 4) wide ranging research on FXS has demonstrated deficits in behavioural inhibition as a core feature of the syndrome (Cornish et al., 2004). In addition, abnormalities have been found in the activation of brain areas during behavioural inhibition tasks and the extent of these abnormalities have been found to be associated with FMRP levels in females with FXS (Menon, Leroux, White, & Reiss, 2004). These findings raise the possibility that gaze aversion, a central feature of the FXS phenotype, may reflect an impairment of the fronto-cortical brain system to override and inhibit the inherent aversion and threat of direct eye contact in contexts that would otherwise be associated with safety. Some evidence exists to suggest that gaze avoidance in FXS reduces as the level of 'threat' in an environment reduces. For example, the current study reported that gaze avoidance behaviours were inhibited when embedded within praise for three individuals. Other studies have noted that gaze avoidance reduces over time in FXS as individuals with FXS become more familiar with another person (e.g., Hall, Lightbody, Huffman, Lazzeroni, & Reiss, 2009). Finally, a recent study by Hall, Lightbody, McCarthy, Parker, and Reiss (2012) has demonstrated that the administration of oxytocin (a hormone that promotes affiliative bonding and inhibits anxiety and associated defensive behaviours) to males with FXS significantly reduced gaze avoidance behaviours and levels of salivary cortisol. Although largely hypothetical, it would be of interest for future research to begin to investigate whether such relations between behavioural inhibition and gaze avoidance exist and serve to underpin gaze aversion in FXS and whether they are amenable to intervention.

The current study has a number of limitations that restrict both its external and internal validity. First, the inter-rater reliability indices for 'fidgeting' behaviours for two participants were poor, and as such the findings reported for this variable must be interpreted with considerable caution. Secondly, only four participants were included in the current study; whilst this is typical of single-case experimental design research, further replication across a wider range of participants is required to establish the generality of these findings (Sidman, 1960). Given that the participants were relatively homogenous in terms of level of intellectual disability, it would be of interest to examine whether similar findings apply to males with FXS with mild and/or severe-profound intellectual disabilities. Third, the number of sessions that could be completed for each individual was restricted by the amount of time available. It is unclear whether the pattern of between-session data would be different had additional sessions been possible or if testing could have continued until differentiation of the data paths appeared. Fourth, there was considerable variability in the between-session data for all participants; which suggests that 'third variables' outside of experimental control were operative and exerted an influence over the current results. Finally, the current study did not control consequences available for target behaviours, and relied solely on antecedent manipulations. As such the data cannot be considered to provide a complete functional analysis of gaze avoidance or other target behaviours (Baer, Wolf, & Risley, 1968).

Despite these limitations, the study has added to the understanding of gaze avoidance in FXS. It seems likely that gaze avoidance in FXS, emerges from the complex interplay between genetic and environmental events. The direct 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).*

This developmental, epigenetic, perspective will be critical in efforts to continue to delineate the nature of gaze avoidance in FXS.

PUBLIC DOMAIN BRIEFING DOCUMENT

Antecedent Influences on Negatively Reinforced Behaviour. An Examination of Person-Environment Interplay

Submitted in partial completion of a Doctorate in Clinical Psychology at the University of Birmingham by Paul Langthorne under the Supervision of Professor Chris Oliver

Background

A literature review was completed as part of the current thesis, which examined the contribution made by variables that alter the 'motivation' for an individual to engage in problem behaviours that are negatively reinforced by escape or avoidance from aversive stimuli. The review presented a model of such behaviours, emphasizing the importance of person-environment interplay. One example of this type of interaction is the interplay between environmental factors (i.e., noise) and aspects of particular phenotypes associated with specific genetic syndromes (i.e., hyperacusis in Williams syndrome). Despite its likely applied importance there is scant research to have investigated such relations however. The empirical study aimed to contribute towards this line of research.

Fragile X syndrome represents the most common inherited form of intellectual and developmental disability and gaze avoidance is considered to form part of the phenotype of this syndrome. The function served by such behaviours (i.e., its interaction with antecedent and consequent environmental conditions) remains poorly understood however, and has typically been inferred by behavioural topography. The current study aimed to further delineate the role

played by gaze avoidance in fragile X syndrome and its relation to both environmental conditions and arousal-related behaviours.

Method

Single-case experimental design methodology was used to examine the influence of systematically manipulating the level of eye contact provided to four boys with FXS whilst completing academic tasks. Using an alternating treatment design, participants were repeatedly exposed to an Eye Contact condition, in which eye contact was provided antecedent, during or consequent to demand presentation or a No Eye Contact condition, in which the same task was presented using minimal eye contact. The influence of this manipulation on both gaze avoidance behaviours and arousal-related variables was then examined. Between-session behavioural data were analysed using Cliff's d statistic, to help determine whether there was a difference between each of the two conditions. Within-session sequential lag analyses were also used to help examine the relationship between particular variables.

Results

Specific types of gaze avoidance behaviours were more likely to occur under Eye Contact conditions, whereas other forms of this behaviour appeared to be unrelated to this manipulation. Data showed that for three participants these specific behaviours appeared to be, at least in part, maintained by negative reinforcement (i.e., were evoked by prompts for eye contact and were followed by a reduction in mutual gaze). Data for the other participant was not consistent with this interpretation and suggest that an alternative mechanism accounts for the heightened probability of these behaviours in Eye Contact conditions. It also appeared that specific forms of

arousal-related behaviours (fidgeting, reported indices of anxiety) were more likely to occur and that yawning was reduced in Eye Contact conditions. This suggests that the high levels of eye contact may have been associated with an increase in arousal. Within-session analyses suggested an association between gaze avoidance behaviours and arousal-related behaviours. For two participants, embedding the delivery of eye contact in praise appeared to be associated with a reduction in gaze avoidance behaviours.

Discussion

The current study has contributed to the literature on gaze avoidance in fragile X syndrome by helping to delineate the extent to which it is influenced by eye contact. Consistent with existing theories of gaze avoidance, evidence was provided to suggest that for some individuals then such behaviours may be maintained by escape from eye contact. However evidence was also provided to suggest the operation of other mechanisms. In addition, all participants presented with forms of gaze avoidance that did not appear to be sensitive to eye contact. The current study also provided evidence to suggest an association between eye contact, increased arousal and gaze avoidance behaviours in fragile X syndrome. Previous research has noted deficits in behavioural inhibition associated with fragile X syndrome. It may be that gaze avoidance in fragile X syndrome, represents a difficulty in the utilisation of fronto-cortical networks to help inhibit the threat amygdale-related response evoked by direct eye contact. It would be of interest for future research to examine whether such postulations could account for gaze avoidance in fragile X syndrome. A number of limitations exist with the current study that may limit its internal and external validity. However, despite these limitations the current study provides an example of phenotype-environment interplay and highlights it applied importance.

APPENDICES

Appendix 1. Data for Forms of Gaze Avoidance.

		Eye Cover		Eye Rub		Turning	
		Eye Contact	No Eye Contact	Eye Contact	No Eye Contact	Eye Contact	No Eye Contact
Terry	Mean (range)	11.23 (3.97-15.45)	3.9 (0.79-10.4)	8.35 (3.17-13.01)	7.94 (0-27.2)	33.75 (16.67-51.22)	43.02 (10.39-85.6)
	Cliff's d	0.83		0.28		-0.22	
Martin	Mean (range)	N/A	N/A	17.73 (9.68-23.33)	7.11 (0-10.17)	27.09 (8.2-38.33)	35.22 (15.25-54.39)
	Cliff's d	N/A		0.92		-0.28	
David	Mean (range)	4.21 (0-16.13)	3.95 (0-10)	12.64 (0-40.32)	17.75 (8.33-28.33)	41.31 (33.33-54.1)	10.89 (6.45-15)
	Cliff's d	-0.16		-0.36		1	
Nick	Mean (range)	1.925 (0-5.77)	1.31 (0-3.33)	0%	0.47 (0-1.89)	31.47 (13.64-50)	17.78 (5.66-34.48)
	Cliff's d	0		-0.25		0.33	

Appendix 2. Data for Forms of Hyperarousal.

		Signs of anxiety		Fidgeting	
		Eye Contact	No Eye Contact	Eye Contact	No Eye Contact
Terry	Mean (range)	23.01 (9.52-40.94)	24.13 (11.2-40.8)	50.24 (38.62-72)	41.02 (24.03-57.02)
	Cliff's d	-0.06		0.33	
Martin	Mean (range)	32.38 (3.23-62.9)	10.74 (1.52-34.33)	11.58 (3.28-30)	19.50 (10.17-28.57)
	Cliff's d	0.60		-0.52	
David	Mean (range)	42.11 (0-85.71)	5.98 (0-20)	20.88 (0-80)	16.91 (8.33-22.58)
	Cliff's d	0.68		-0.52	
Nick	Mean (range)	19.44 (0-33.33)	0.94 (0-3.77)	45.41 (16.67-77.27)	41.17 (30.19-48.28)
	Cliff's d	0.58		-0.17	

Appendix 3. Data for Escape-Related Behaviour.

		Elopement		Property destruction		Tangential social initiations		Requests to terminate task	
		Eye Contact	No Eye Contact	Eye Contact	No Eye Contact	Eye Contact	No Eye Contact	Eye Contact	No Eye Contact
Terry	Mean (range)	0.54 (0-2.44)	0.49 (0-1.65)	N/A	N/A	14.85 (3.2-28.28)	3.46 (0-7.2)	N/A	N/A
	Cliff's d	0		N/A		0.83		N/A	
Martin	Mean (range)	1.33(0-6.67)	5.62 (0-17.57)	0.65 (0-3.23)	21.14 (0-44.59)	N/A	N/A	N/A	N/A
	Cliff's d	-0.36		-0.52		N/A		N/A	
David	Mean (range)	0.32 (0-1.59)	0	N/A	N/A	N/A	N/A	N/A	N/A
	Cliff's d	0.20		N/A		N/A		N/A	
Nick	Mean (range)	39.44 (17.46-82.35)	28.41 (0-62.16)	15.66 (0-33.33)	0.83 (0-3.33)	N/A	N/A	3.44 (0-5.77)	9.4 (0-16.67)
	Cliff's d	0.33		0.58		N/A		-0.58	

Appendix 4. Data for Additional Variables of Interest.

		Yawning		Stereotypical behaviour	
		Eye Contact	No Eye Contact	Eye Contact	No Eye Contact
Terry	Mean (range)	1.56 (0-4)	5.2 (0-12.4)	19.49 (8.28-39.37)	9.45 (0-39.2)
	Cliff's d	-0.22		0.67	
Martin	Mean (range)	0	1.81 (0-7.46)	42.87 (26.67-54.84)	16.67 (8.11-25.37)
	Cliff's d	-0.4		1	
David	Mean (range)	2.6 (0-6.56)	9.99 (6.67-14.52)	N/A	N/A
	Cliff's d	-1		N/A	
Nick	Mean (range)	0	2.36 (0-9.43)	N/A	N/A
	Cliff's d	-0.25		N/A	

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