An Investigation of Physiological Arousal in Children with Autism and Co-morbid Challenging Behaviour

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Summary

Challenging behaviour is prevalent among individuals with autism spectrum disorders (ASD). While operant theory has allowed for the development of effective behavioural interventions for challenging behaviours such as self-injury and stereotypy, the aetiology of such behaviours remains largely unknown. An abundance of theories exist that posit that these behaviours may be the result of physiological dysregulation or atypical physiological processes. However, there is currently a limited body of research empirically examining the relationship between physiological activity and engagement in challenging behaviour. The purpose of the current program of research was to examine the contribution of psychophysiological measurement to our understanding and assessment of challenging behaviour in ASD.

Chapter 1 provides an overview of the extant literature pertaining to challenging behaviour in ASD, physiological functioning among those with the disorder, and the theoretical and empirical work suggestive of a relation between physiological activity and engagement in challenging behaviours by those with ASD.

Chapter 2 presents a systematic review of physiological reactivity to sensory, social and emotional, and stressor stimuli among individuals with ASD. The aim of this review was to examine whether physiological reactivity to stimuli is atypical in this population and whether differences in physiological reactions to stimuli may contribute to the atypical behaviours associated with autism. The results reveal that previous research has yielded inconsistent findings across studies reviewed with individuals with autism responding differently to typically developing controls on 78.6%, 66.7% and 71.4% of sensory, social and emotional, and stressor stimuli respectively. It was found that the disparity in outcomes was not attributable to the methodological quality of included studies and that extant research suggests that physiological reactivity is associated with a number of

behaviours and psychological variables related to autism.

Chapter 3 investigates cortisol levels among children and adolescents with autism (*n*=52) as compared to a matched sample of typically developing peers. Further, the relationship between levels of stress- parent-reported and physiological-and challenging behaviour was examined for 61 youths with autism. While not statistically significant, there was a trend towards higher levels of stress among participants diagnosed with autism. Cortisol levels were not correlated with participants' self-injurious and aggressive behaviours. However, it was found that participants who engaged in high levels of stereotypy had higher cortisol levels (large effect size) than participants who engaged in little or no stereotypy.

Chapter 4 examines the relationship between heart rate and motor stereotypy among five children and adolescents with ASD. Our analysis did not suggest that physiological arousal functioned as an antecedent to, or reinforcer for, four of the five participants' stereotypy. For one participant, stereotypy consistently resulted in heart rate decreases that may have acted as negative reinforcement for this behaviour. The examination of heart rate levels during various mood states in this study suggested that participants presented with an atypical physiological response, suggestive of physiological blunting, to stressors encountered in their natural environment during the recording period.

Chapter 5 assesses the contribution of heart rate and electrodermal activity to the experimental functional analysis of repetitive, stereotyped behaviour among six children and adolescents diagnosed with autism. Our analysis revealed little association between physiological activity and engagement in repetitive, stereotyped behaviours for these participants. However, similar to the findings reported in Chapter 4, an apparent blunted physiological response to stressors encountered was noted for a number of the participants.

The results of the current research program are discussed in Chapter 6 in relation to their implications for behaviour analysts' understanding, assessment, and treatment of challenging behaviour among individuals with autism. Our results suggest that psychophysiological assessments among persons with autism can offer a valuable contribution to behaviour analysts' understanding of challenging behaviour and the experiences and wellbeing of persons with autism. However, it was found that these assessments may not yield useful data for all individuals with autism. Future research must therefore establish the behaviours, individuals, or conditions for which these assessments are most useful. Additional directions for further research on the experience of stress among persons with autism, physiological blunting among this population, potential physiological subgroups of autism, and the inclusion of lower functioning individuals with autism in psychophysiological research studies, are also described.

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List of Abbreviations

ABA:	Applied behaviour analysis
ACTH:	Adrenocorticotropic hormone
ADHD:	Attention deficit hyperactivity disorder
ADOS:	Autism Diagnostic Observation Schedule
ANS:	Autonomic nervous system
ASD:	Autism spectrum disorder
BP:	Blood pressure
BPI-01:	The Behavior Problems Inventory
BPM:	Beats per minute
CARS	Childhood Autism Rating Scale
DSM	The Diagnostic and Statistical Manual of Mental Disorders
EDA:	Electrodermal activity
HR:	Heart rate
HRV:	Heart rate variability
LHPA:	Limbic-hypothalamic-pituitary-adrenal
PDS:	The Pubertal Development Scale
PNS:	Parasympathetic nervous system
PR:	Physiological reactivity
RBS-R:	Repetitive Behavior Scale-Revised
SCQ:	Social Communication Questionnaire
SCS:	Salimetrics Children's swab
SIB:	Self-injurious behaviour
SNS:	Sympathetic nervous system
SSS:	The Stress Survey Schedule for persons with autism and other developmental disabilities
Ųg/dl:	Micrograms per decilitre
ųS:	MicroSiemens

Chapter 1

General Introduction

"The physiologist of the future will tell us all that can be known about what is happening inside the behaving organism…What he discovers cannot invalidate the laws of a science of behaviour, but it will make the picture of human action more nearly complete"

(Skinner, 1974, p. 215)

Challenging Behaviour in Autism Spectrum Disorder

Challenging behaviour is behaviour "of such intensity, frequency or duration that the physical safety of the person or others is likely to be placed in serious jeopardy, or behaviour which is likely to seriously limit use of, or result in the person being denied access to, ordinary community facilities" (Emerson, 2001, p.3). Challenging behaviour is a significant problem for the parents, educators, and service providers of individuals with autism spectrum disorder (ASD); Murphy, Healy, and Leader (2009) investigated the prevalence of challenging behaviours among a sample of 157 Irish children diagnosed with ASD. It was found that 82% of the sample engaged in at least one form of challenging behaviour while 32.5% presented with self-injurious behaviour (SIB), aggression and stereotypy. Similarly, Jang et al. (2011) found that among a sample of 84 children with ASD, 94% of the sample presented with some form of challenging behaviour. Studies have found that challenging behaviour persists throughout the lifetime, and those displaying challenging behaviour earlier in life tend to engage in such behaviours even when they are older (Murphy et al., 2005).

Challenging behaviour negatively impacts the social development and social interactions, quality of life, and the education and learning, of individuals diagnosed with developmental disabilities in a myriad of ways. Such behaviours are socially stigmatising and may hamper integration in the community or typical education settings, hinder social interaction with peers or adults, or lead to removal from the community or home and

placement in a restrictive service setting (Cunningham & Schreibman, 2008; Symons, 1995; Thompson, Egli, Symons, & Delaney, 1994). Matson and Boisjoli (2009) found that, among a sample of individuals with intellectual disabilities and challenging behaviour, the topography, frequency, and intensity of challenging behaviours were risk factors for chemical or mechanical restraint. SIB has been found to be associated with a number of negative social consequences, such as residential placement, experience of aversive interventions, and the use of restraint, in addition to resulting in significant health or medical issues, such as tissue damage or loss, retinal detachment, infection, and cauliflower ear (Favell et al., 1982; Mace & Mauk, 1999). Stereotypy has been shown to detrimentally affect learning and other positive behaviours. Longer response latencies to sensory stimuli (Lovaas, Litrownik, & Mann, 1971), impaired play behaviours (Koegel, Firestone, Kramme, & Dunlap, 1974), and the failure to master simple discriminations (Koegel & Covert, 1972), have all been evidenced among individuals who engage in stereotyped behaviour. Challenging behaviour does not solely affect the individual who engages in it, it can also profoundly affect the lives of parents or other caregivers; several studies have identified a link between challenging behaviours and parental, teacher, or caregiver stress (Davis & Carter, 2008; Hastings, 2002; Herring et al., 2006; Lecavalier, Leone, & Wiltz, 2005; Smyth, Healy, & Lydon, 2015). Furthermore, the financial costs of caring for an individual who engages in challenging behaviour are considerable (Thompson et al., 1994).

The relationship between challenging behaviour and reduced quality of life makes research into its cause a high priority. The proposed research will investigate the relationship between physiological activity and two of the most common and chronic forms of challenging behaviour among individuals with ASD; SIB and stereotypy (Murphy et al., 2005; Murphy et al., 2009).

Self-injurious Behaviour

SIB has been defined as any behaviour that leads to the physical harm of one's own body (Tate & Baroff, 1966). Such behaviour is highly prevalent among individuals diagnosed with ASD, with several studies producing incidence estimations of over 50% (Baghdadli, Pascal, Grisi, & Aussilloux, 2003; Duerden et al., 2012; Richards, Oliver, Nelson, & Moss, 2012). Myriad topographies of SIB have been described in the literature including behaviours such as head banging, biting, hand mouthing, self-hitting, pica, rumination, scratching, hair pulling, eye-poking, ear-poking, skin-picking, pinching, bruxism, and choking (Iwata et al., 1994a; Kahng, Iwata, & Lewin, 2002; Lydon, Healy, Moran, & Foody, 2015a). Identified risk factors for SIB include age, autism severity, atypical sensory processing, level of cognitive functioning, language ability, level of adaptive functioning, insistence on sameness or resistance to change, impulsivity, hyperactivity, negative affect, engagement in stereotypy or restricted repetitive behaviour, poor health, and perinatal conditions (Baghdadli et al., 2003; Duerden et al., 2012; Jang et al., 2011; McTiernan, Leader, Healy, & Mannion, 2011; Petty, Bacarese-Hamilton, Davies, & Oliver, 2014; Richards et al., 2012; Richman et al., 2013).

Often considered the most serious co-morbid challenging behaviour among individuals with autism (Devine, 2013), the aetiology of SIB has received much research attention but is still largely unknown (Duerden et al., 2012; Forgeot d'Arc, Dawson, Soulières, & Mottron, 2012; Iwata, Dorsey, Slifer, Bauman, & Richman, 1994b; Kurtz, Chin, Huete, & Cataldo, 2012; Sterling, McLaughlin, & King, 2011; Waters & Healy, 2012). Symons (1995; Symons & Thompson, 1997) divided existent theories regarding the aetiology of SIB into seven categories: 1) psychodynamic theories; 2) organic or biologically-based theories; 3) developmental theories; 4) side effects of minor illness theories; 5) self-stimulation theories; 6) learned behaviour theories, and 7) neurochemical theories. The degree of evidence supporting each theory varies; for example, there are no

experimental studies that support the suggestions of psychodynamic theories and treatments based on these have been found to be ineffective for reducing SIB (Symons, 1995). Similarly, there is limited research to support developmental theories which suggest that SIB is a common behaviour among young typically developing children and that the SIB of individuals with autism or other developmental disabilities represents a delay in development or the continuation of these early motor behaviours. Instead, research has shown that there is little correspondence between the self-injury emitted by young children and that of individuals with developmental disabilities and has failed to identify a developmental function of such behaviours (Symons, 1995).

In contrast, greater support for biological and neurochemical theories of SIB has been published. Most commonly, neurochemical theories implicate altered dopaminergic functioning (Breese et al., 1984a; Breese et al., 1984b; Lloyd et al., 1981; Schroeder & Tessel, 1994), addiction to opiate hormones which prompts SIB in order to avoid the effects of opiate withdrawal (Sandman, 1990; Sandman et al., 1993; Sandman & Hetrick, 1995; Thompson, Symons, Delaney, & England, 1995), or altered serotonergic functioning (Crowell et al., 2008; Kolevzon et al., 2010; Schroeder & Tessel, 1994). Altered brain structures have also been implicated; for example, Wolff and colleagues (2013) identified a positive correlation between bilateral caudate nuclei volume and self-injury among 30 boys diagnosed with autism.

The current research program is founded primarily on the well-supported operant theories of SIB which suggest that SIB can be maintained by environmental consequences through mechanisms of social positive reinforcement or social negative reinforcement (Hanley, Iwata, & McCord, 2003; Iwata et al., 1994a; Kurtz et al., 2003). Several reviews of the literature also conclude that SIB is frequently identified as automatically reinforced during functional analyses (Hanley et al., 2003; Iwata et al., 1994a; Kurtz et al., 2003). However, to my knowledge, only two papers (Barrera, Violo, & Graver, 2007; Hall,

Hammond, & Hustyi, 2013) have succeeded in identifying the specific source of automatic reinforcement, automatic negative reinforcement in the case of Barrera et al. and automatic positive reinforcement in the case of Hall et al., responsible for the maintenance of the SIB of individuals diagnosed with a variety of developmental disabilities. Behavioural researchers have also suggested that SIB may serve a communicative function for individuals who are not vocal or who cannot communicate effectively (Carr & Durand, 1985; Twachtman-Cullen, 2006). The veracity of behavioural theories of SIB is evidenced by the myriad of studies that have demonstrated the efficacy of function-based behavioural interventions for reducing or eliminating SIB (e.g., Campbell, 2003; Heyvaert, Saenen, Campbell, Maes, & Onghena, 2014; Kahng et al., 2002). Carr (1977) reviewed evidence for the various etiological theories of SIB and concluded that such behaviours are likely to be multiply-controlled and that there is a low likelihood that a single explanatory factor will be identified. His proposition that each case of SIB is idiosyncratic and that a comprehensive assessment is necessary to determine the factors responsible for an individual's SIB and its maintenance has been reiterated many times since (Guess & Carr, 1991; Iwata et al., 1994b; Mace & Mauk, 1999; MacLean, Stone, & Brown, 1994; Sterling et al., 2011; Symons, Sperry, Dropik, & Bodfish, 2005).

Stereotypy

Restricted, repetitive, and stereotyped behaviours, most commonly referred to as stereotypy in the literature, are one of the core features of ASD (American Psychiatric Association, 2013). Stereotyped behaviour is not unique to ASD, however there is evidence that it is more frequent and severe among individuals with ASD than among individuals diagnosed with intellectual disabilities, other developmental disabilities, or typically developing individuals (Bodfish, Symons, Parker, & Lewis, 2000; Cervantes, Matson, Williams, & Jang, 2014; Goldman et al., 2009; MacDonald et al., 2007; Matson et al., 1996). Campbell and colleagues (1990) examined stereotyped movements among

children with ASD and found that all but one child (99.5%) were rated as having at least mild stereotyped behaviour. Murphy et al. (2009) found that 27.3% of their sample presented with stereotypy alone, while 11% presented with stereotypy and SIB, and a further 32.5% presented with stereotypy, SIB, and aggressive behaviours. A wide variety of topographies of stereotypy have been reported in the literature including body rocking, head tilting, object movement or mouthing, hand or finger movement, facial movements, hand gazing, tip toe walking, and covering eyes or ears (Lydon et al., 2015a; Symons et al., 2004). Identified risk factors for stereotypy include autism severity, cognitive impairment, deficits in adaptive functioning, and hyperactivity (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Goldman et al., 2009; Matson, Kiely, & Bamburg, 1997; McTiernan et al., 2011). Cervantes et al. (2014) found that children with ASD and typical IQ (IQ of greater than 70) displayed more frequent stereotypy than children with ASD and low IQ (IQ of 70 or less).

Stereotypy and SIB have been shown to be interrelated; studies have described "self-stimulatory", or stereotyped, SIB and others have suggested that high-intensity stereotyped behaviours can become self-injurious (Duerden et al., 2012; Guess & Carr, 1991; Nyhan, 1994; Sterling et al., 2011). Oliver and colleagues (2012) found that repetitive behaviour was a risk factor for SIB and other challenging behaviours among a sample of 943 children diagnosed with intellectual disabilities; participants were 16 times more likely to engage in SIB if they presented with high frequency repetitive behaviours. Richman and colleagues (2013) found that, among a sample of 617 individuals with ASD, stereotyped behaviour was highly predictive of SIB. Such findings suggest that it is possible that these behaviours share a common aetiological component.

A multitude of theories have attempted to explain engagement in stereotyped behaviour by individuals with autism. Ornitz (1974) suggested that stereotypy is related to the faulty modulation of sensory input such that children with ASD engage in stereotypy to

produce additional kinaesthetic stimulation and a greater understanding of their environment. Other scholars have proposed the homeostatic theory of stereotypy that suggests that there exists an optimal level of sensory stimulation and that stereotypy serves a homeostatic function by increasing or decreasing sensory stimulation in under- or over-arousing environments (Lewis & Baumeister, 1982; Zentall & Zentall, 1983). Experts have also suggested that stereotypy is non-functional and merely the result of physiological pathology; a number of studies have identified an association between atypical brain structure and stereotypy among individuals with autism (e.g., Langen et al., 2014; Pierce & Courchesne, 2001; Wolff et al., 2013). As with SIB, a variety of neurochemicals have also been implicated in the emergence of stereotyped behaviours; dopamine has been implicated in the emergence of stereotypy due to demonstrations that dopamine agonists reduce this behaviour or that dopamine receptor lesions in animals increase the likelihood of stereotyped behaviour (Lewis, Gluck, Bodfish, Beauchamp, & Mailman, 1996; Ridley, Baker, & Scraggs, 1979). The reductions in stereotypy following the administration of serotonin uptake inhibitors (Garber, McGonigle, Slomka, & Monteverde, 1992; Lewis, Bodfish, Powell, & Golden, 1995) or opiate antagonists (Cazzullo et al., 1999; Smith, Gupta, & Smith, 1995) have also led researchers to postulate a relationship between these neurochemicals and stereotypy.

Of primary relevance to the current work is the operant conceptualisation of stereotypy. The behavioural paradigm proposes that stereotypy is an operant behaviour that is maintained by the reinforcing consequences that follow its occurrence (Lovaas, Newsom, & Hickman, 1987; Rapp & Vollmer, 2005a). This theory has been supported by applied behaviour analytic studies that have identified the behavioural function of stereotypy and used this information to develop effective, function-based treatments (see Campbell, 2003; Heyvaert et al., 2014; Mulligan, Healy, Lydon, Moran, & Foody, 2014 for reviews of this literature). Several studies have identified sources of social positive

reinforcement or social negative reinforcement (Durand & Carr, 1987; Hanley et al., 2003; Kennedy, Meyer, Knowles, & Shukla, 2000; Rehfeldt & Chambers, 2002), automatic positive reinforcement (e.g. Piazza, Adelinis, Hanley, Goh, & Delia, 2000; Rapp, Dozier, Carr, Patel, & Enloe, 2000), and automatic negative reinforcement (e.g. Tang, Kennedy, Koppekin, & Carruso, 2002) responsible for maintaining the stereotyped behaviour of participants. However, most frequently, stereotyped behaviours are identified as being automatically reinforced (Cunningham & Schreibman, 2008). The common usage of behaviour modification procedures, in lieu of function-based behavioural interventions, to treat automatically reinforced stereotypy suggests that the classification of these behaviours as "automatically reinforced", and the non-identification of the specific source of reinforcement for the behaviour, is often not helpful with the development of function-based behavioural interventions (Rapp & Vollmer, 2005a).

Physiological Activity in Autism Spectrum Disorder

Researchers have suggested that challenging behaviours such as stereotypy and SIB may result from underlying dysregulation in physiological functioning among individuals with ASD. A number of physiological systems have been identified as potentially impaired in this population. Of primary relevance to the current research is the functioning of the autonomic nervous system (ANS) and the limbic-hypothalamic-pituitary-adrenal (LHPA) axis in ASD.

The Autonomic Nervous System

The ANS consists of two complementary branches that work together to preserve homeostasis in the body: the parasympathetic nervous system (PNS) and the sympathetic nervous system (SNS). The primary function of the SNS is to activate, or mobilise, the organism in response to stress or threat, often referred to as the "fight or flight" response (Lovallo, 2005). Conversely, the PNS is responsible for calming the organism, promoting the restoration of homeostasis, and allowing for the resumption of normal processes such

as the absorption of nutrients, conservation of energy, and occurrence of growth (Turner, 1994). Activation of the ANS in response to stress or stimulation typically occurs quickly and the level of activation can be readily assessed through measures of heart rate (HR), heart rate variability (HRV), blood pressure (BP), and electrodermal activity (EDA) (Boyce & Ellis, 2005; Lovallo, 2005). A delicate balance exists between the PNS and SNS that is essential for physical and mental wellbeing; research has demonstrated that autonomic dysfunction comprising the excessive activation of one system over the other results in physical ill health (Thayer & Lane, 2007) and is implicated in a number of psychological disorders including anxiety disorders, schizophrenia, and attention deficit hyperactivity disorder (ADHD; Klusek, Roberts, & Losh, 2015).

Given that the ANS has been described as an essential component of behavioural regulation (Porges, 2001), it is perhaps unsurprising that theories which posit the dysregulation of the ANS among those with ASD abound. Initial theories suggested that individuals with ASD experience hyper-arousal of the ANS (Hutt, Hutt, Lee, & Ounsted, 1964). However, decades of subsequent experimental investigations of autonomic activity, and autonomic reactivity, in ASD have yielded inconsistent findings regarding the existence of such deficits in this population (Benevides & Lane, 2013; Rogers & Ozonoff, 2005; Sugarman, Garrison, & Williford, 2013). Significant differences identified in research studies may represent true differences in ANS functioning in this population or may be related to experimental protocols, participant characteristics, participant engagement in restricted, repetitive behaviours, and poor ecological validity (Benevides & Lane, 2013; Sugarman et al., 2013). The understanding of ANS functioning in ASD is further complicated by findings of subgroups of persons with ASD experiencing either physiological hyper-arousal or physiological hypo-arousal (e.g., Hirstein, Iverson, & Ramachandran, 2001; Schoen, Miller, Brett-Green, & Hepburn, 2008); the existence of such individuals experiencing opposite extremes of baseline physiological arousal and

different patterns of physiological reactivity (PR) likely contributes to the inconsistent findings observed in this area of research.

The Limbic-Hypothalmic-Pituitary-Adrenal Axis

The LHPA axis is a key component of the neuroendocrine system that regulates the physiological response to stress. The activation of the LHPA axis in response to stress results in the production of adrenocorticotropic hormone (ACTH) which then stimulates production of cortisol, a stress hormone (Kalat, 2004). Cortisol can be extracted from a number of biological fluids including saliva, blood, and urine. In contrast to the ANS, LHPA activation in response to stress is typically slower. The release of cortisol impacts upon the cardiovascular system, metabolic functioning, and immunological functioning (Sapolsky et al., 2000). While cortisol reactivity to stressors is adaptive, prolonged elevation of cortisol levels has been linked to negative health outcomes (Kalat, 2004). Dysfunction of the LHPA axis has been implicated in a number of disorders including post-traumatic stress disorder and other anxiety disorders (Pitman et al., 2012; Staufenbiel, Penninx, Spijker, Elzinga, & van Rossum, 2013), depression (Burke, Davis, Otte, & Mohr, 2005), and ADHD (Hastings, Fortier, Utendale, Simard, & Robaey, 2009).

Cortisol has been suggested as a potential biomarker for the diagnosis of ASD (Yang et al., 2015). However, extensive investigation of LHPA axis functioning in ASD has resulted in conflicting findings (see Nuske, Vivanti, & Dissanayake, 2014; Taylor & Corbett, 2014; Tordjman et al., 2014 for reviews) regarding baseline cortisol levels, cortisol diurnal patterns, cortisol variability, and cortisol reactivity in response to stressors. A review by Taylor and Corbett (2014) suggested that LHPA axis dysfunction exists for lower-functioning individuals with ASD only, and that high-functioning ASD is not associated with the dysregulation of cortisol rhythms. Recent research (e.g., Tordjman et al., 2014; Putnam, Lopata, Thomeer, Volker, & Rodgers, 2015) examining diurnal salivary cortisol levels among high-functioning and low-functioning individuals with ASD has

corroborated this suggestion by identifying differences in LHPA axis functioning by level of cognitive functioning.

Theories linking Physiological Activity and Challenging Behaviour

Theories that associate challenging behaviour, such as SIB and stereotypy, and physiological functioning first emerged in the 1960's (Hutt et al., 1964; Hutt & Hutt, 1968). Hutt and colleagues' (1964) observations of increased EEG activation among children with ASD, more frequent stereotypy in response to more stimulating environments, and a correlation between EEG activation and stereotypy prompted them to develop an arousal modulation theory of autism. This theory proposed that individuals with ASD experience physiological hyper-arousal that leads to their increased behavioural reactivity to environmental stimuli and their avoidance of social interaction and novel situations. Hyper-arousal involves excessive SNS activity and significantly elevated physiological arousal, manifested through increased HR and respiration, which interferes with cognitive processing and can lead to feelings of anxiety, hyper-vigilance, and aggression towards others (Ogden & Minton, 2000). Hutt and Hutt (1968) further noted that increased rates of stereotypy were associated with situations, such as novel environments, interactions with strangers, and the presence of unfamiliar objects, which would be expected to result in increased physiological arousal, and that participants with ASD presented with EEG levels indicative of baseline hyper-arousal. They suggested that the repetitive behaviours characteristic of ASD might be understood as monotonous stimulation that has an arousal reducing effect. Later researchers, such as Dawson (1991) who suggested that children with ASD showed a failure to habituate to sensory stimuli which leads to uncomfortably elevated levels of physiological arousal, also reiterated this theory. Conversely, physiological hypo-arousal which involves excessive PNS activity which leads to reduced physiological arousal, including lowered HR and respiration, the slowing of motor responses, feelings of numbness or social isolation, and disruptions in

cognitive and affective processing, has also been suggested as a potential explanation for the abnormal behaviours common among individuals with ASD (Ogden & Minton, 2000; Schoen et al., 2008). For example, Edelson (1984) proposed that individuals with ASD have an increased sensory threshold that leads to feelings of hypo-arousal and causes them to engage in self-stimulatory behaviours.

It may be the case however, that both extremes of arousal exist among individuals with ASD; Schoen and colleagues (2008) identified two distinct types of participants among their sample of children with high-functioning autism or Asperger's syndrome. First, a low-arousal group who were physiologically hypo-aroused, did not respond physiologically to the stimuli presented, and were slow to react. Second, a high-arousal group who were shown to be physiologically hyper-aroused, showed increased PR to stimuli, and responded quickly to experimental stimuli. Earlier, Hirstein and colleagues (2001) had identified similar low- and high-arousal groups among their sample of participants with ASD. Ogden and Minton (2000) also suggested that individuals experiencing dysregulated physiological arousal may consistently experience one extreme of arousal, e.g., hypo- or hyper-arousal, or may fluctuate between the two states. Research has evidenced both physiological hyper-arousal (e.g., Chang et al., 2012; Goodwin et al., 2006; Palkowitz & Wiesenfeld, 1980) and physiological hypo-arousal (e.g., Graveling & Brooke, 1978; Hubert, Wicker, Monfardini, & Deruelle, 2009) among individuals with ASD. However, several studies have also found that levels of physiological activity among those with ASD do not differ significantly from those of typically developing controls (e.g., Naber et al., 2007; Lanni, Schupp, Simon, & Corbett, 2012; Levine, Sheinkopf, Pescosolido, Rodino, Elia, & Lester, 2012).

Romanczyk (1986) appears to have been among the first to recognise the potential contribution of physiological data to the behavioural assessment or analysis of challenging behaviour as he emphasised that "acknowledgement must be given to the importance of

physiological variables and the strong potential that biological (organic) factors are of great importance in producing a comprehensive analysis of self-injurious behaviour" (p. 42). Romanczyk was instrumental in the development of an operant-respondent model of SIB that highlights physiological arousal as an important contributory factor in the elicitation and maintenance of SIB (Romanczyk, 1986; Romanczyk & Matthews, 1998; Romanczyk, Lockshin, & O'Connor, 1992). In this theory, SIB is seen as an aversive, painful event. SIB is initially a respondent behaviour that is elicited by uncomfortable or painful levels of physiological arousal, which result from environmental stressors. Certain environmental variables may become associated with high levels of physiological arousal and may come to elicit SIB even when elevated physiological arousal is absent. Over time, environmental events, or social variables, can also come to maintain SIB through operant conditioning processes. In this way, SIB becomes an increasingly functional behaviour for the individual and is likely to persist in the absence of intervention.

Guess and Carr (1991) proposed a three-level model of stereotypy and SIB that highlights the potential influence of endogenous factors, such as development or maturation, physiological abnormalities, or sensory impairment, and exogenous factors, such as external reinforcement, environmental stimulation, or environmental stress, on these behaviours. The first level of this model emphasises that these behaviours are related to biological processes and internal regulation and that they are common, and developmentally appropriate, for very young children but that their emergence is delayed among children with developmental disabilities. The second level describes how these behaviours are employed, by individuals with developmental disabilities, in response to environmental stimulation to maintain internal homeostasis or an optimal level of stimulation. The third level details how such behaviours, originally related to internal biological functioning, can subsequently develop operant functions and may be employed to elicit positive or negative reinforcement from others by individuals with developmental

disabilities. The authors claim that while the transition from one level to the next may appear logical, that these levels are not completely independent or hierarchical. Instead, each level may act together, in various combinations, to elicit challenging behaviour, or the controlling level may differ depending on changes in internal or external events.

Haines and colleagues (1995) proposed that SIB functions to reduce, or offer relief from, internal tension, or other unpleasant internal states. They studied a sample of typically developing individuals who either engaged in self-mutilation, which included behaviours such as cutting, skin scratching, and hitting, or no self-mutilation. Among individuals who engaged in self-mutilation, psychophysiological arousal, which encompasses both physiological arousal and feelings of distress, increased progressively throughout a self-cutting imagery script as the scene and precursor behaviours were described and decreased once the decision to engage in self-mutilation was made and the self-cutting occurred. Arousal remained low following the occurrence of the behaviour. Such tension reduction was not observed among participants who did not engage in self-mutilation. Brain, Haines, and Williams (1998) replicated these results and showed once again that individuals who engaged in self-mutilation did not differ from those who did not self-mutilate in response to neutral, control events but experienced increased psychophysiological arousal in response to self-mutilation imagery which only decreased following the occurrence of self-mutilation. The authors propose that SIB may be understood as a coping strategy, employed during periods of increasingly aversive psychophysiological arousal, which is reinforced and maintained by reductions in psychophysiological arousal and co-occurring feelings of relaxation. Barrera et al. (2007) put forward a similar account of SIB, suggesting that among individuals with developmental disabilities SIB is a negatively reinforced behaviour that results in the termination, or escape from, uncomfortably elevated levels of physiological arousal. These

theories posit that the effectiveness of self-injurious behaviours at reducing physiological arousal leads to compulsion to engage in the behaviours and their high rate of occcurrence.

It has also been suggested that stress may play a role in the development or emergence of challenging behaviours such as SIB and stereotypy. Groden, Cautela, Prince, and Berryman (1994) suggested that individuals with ASD have a unique vulnerability to stress and anxiety due to the impairments commonly associated with the disorder such as social and communication deficits, co-occuring cognitive impairment, sensory sensitivity, and the need for sameness. Groden et al. suggest that these variables may contribute to heightened stress and anxiety, reflected in physiological activation, in this population and that challenging behaviours may be precipitated by stressors, may serve as faulty coping strategies, and for some persons may function to reduce physiological arousal. This suggestion is supported by findings that significant stress leads to the release of multiple neurochemicals such as cortisol, epinephrine, norepinephrine, vasopressin, oxytocin, and opioids, several of which have been implicated in the development of SIB (Yates, 2004). Furthermore, personal reports from high-functioning individuals (e.g., Grandin, 2006) often emphasise feelings of extreme stress or anxiety and describe how such internal tension affects overt behaviour lending credence to such theories.

In a series of papers, Langthorne and colleagues (Langthorne, McGill, & MacLean, 2008; Langthorne, McGill, & Oliver, 2014; Langthorne, McGill, & O'Reilly, 2007) have also described the importance of the physiological context in which challenging behaviour occurs and criticised the "historical divide between the biological and behavioural sciences" (Langthorne et al., 2014, p.139). In these papers, the authors have posited that genetic, neurobiological, and physiological variables may serve as motivating operations for engagement in various challenging behaviours in the developmental disability population. Their theory suggests that physiological state(s) may alter the reinforcing value of stimuli or events and impact upon behaviour in this way. While the authors focus

primarily on discrete physiological and biological states (e.g., genetic syndromes, otitis media, sleep, psychiatric conditions, allergies etc.) in their papers, this theory may also be relevant to the study of the relation between physiological activity or physiological activation and engagement in challenging behaviour among persons with ASD. Given suggestions of chronic physiological hypo- and hyper-arousal in this population, these physiological states may act as motivating operations and continually impact upon behaviour and the reinforcing value of stimuli. For example, this theory suggests that low levels of physiological activation (i.e., physiological hypo-arousal) could exert behaviour-altering effects by altering the reinforcing value of increases in physiological arousal, thereby increasing the likelihood of engagement in behaviours that result in increases in physiological activity (e.g., vigorous motor stereotypy). Similarly, high levels of physiological activation (i.e., physiological hyper-arousal) could exert behavioural-altering effects by altering the reinforcing value of decreases in physiological activity and increasing an individual's motivation to engage in behaviours that function to reduce physiological arousal (e.g., SIB or stereotypy).

Lydon, Healy and Dwyer (2013) proposed that challenging behaviours, such as stereotypy and SIB, may function to allow access to a preferred state of elevated physiological arousal. It has been previously suggested that, contrary to the commonly accepted belief that high arousal is aversive and uncomfortable, some individuals may enjoy the feelings and bodily sensations associated with elevated arousal (Svebak & Stoyva, 1980). In this way, challenging behaviours exhibited by individuals with ASD, which have been seen to lead to increased physiological arousal for at least some participants in several studies (Freeman, Horner, & Reichle, 1999; Hirstein et al., 2001; Lewis et al., 1984; Lydon et al., 2013; Sroufe, Stuecher, & Stutzer, 1973; Willemsen-Swinkels, Buitelaar, Dekker, & van Engeland, 1998; Young & Clements, 1979), may function to increase physiological arousal or to maintain already elevated physiological

arousal by individuals who perceive increased physiological arousal as a pleasant internal state. This suggestion is not unique; Eysenck (1997) previously put forward a stimulation-seeking theory of anti-social behaviour which suggests that engagement in certain behaviours may be motivated by physiological deprivation (i.e., hypo-arousal) and reinforced by the increases in physiological arousal produced. Lydon et al.'s theory is also supported by findings of reduced stereotypy and SIB following physical exercise among persons with ASD, particularly following vigorous exercise (Elliott, Dobbin, Rose, & Soper, 1994; Kern, Koegel, & Dunlap, 1984; Lang et al., 2010; Rosenthal-Malek & Mitchell, 1997). Lang and colleagues (2010) suggested that physical exercise, which leads to increases in physiological arousal, may provide the same stimulation as stereotyped behaviours and eliminate the establishing operations for such behaviours. Given that stereotypy and SIB often occur at high frequencies, and that resulting increases in physiological arousal have been evidenced, it is certainly plausible to suggest that physiological arousal may be reinforcing for certain individuals.

Next, Sugarman and colleagues (2014) put forward the Autonomic Dysregulation Theory of Autism. This theory, an extension of the arousal modulation theory first described by Hutt et al. (1964), suggests that impairments in autonomic regulation may underlie the majority of atypical and challenging behaviours observed among individuals with ASD. It suggests that the restricted, repetitive behaviours characteristic of autism may be viewed as displacement behaviours which subsequently may be viewed as an attempt to decrease stress, or high levels of arousal, and to restore homeostasis. The authors interpret the elevated rates of anxiety among individuals with ASD, findings of elevated baseline arousal, and the prevalence of co-morbid sleep disorders, gastrointestinal issues, and tic disorders, as further evidence of autonomic dysregulation. The authors do not pinpoint the exact defect, or deficit, in autonomic regulation, suggesting that further research is needed to identify and elucidate this, but suggest that it results in the social impairments, sensory

issues, and anxiety issues that are pervasive in ASD. The authors view restricted repetitive behaviours, avoidance of sensory stimuli, and insistence on sameness, as serving de-arousal functions and attempts to avoid further increases in arousal.

Most recently, Tordjman and colleagues (2015) have proposed autism to be a disorder of dysregulated biological and behavioural rhythms. The authors highlight a growing body of research suggestive of disordered circadian rhythms among persons with autism, perceptible through the assessment of cortisol circadian rhythms or melatonin secretion. The physiological continuity, and absence of physiological variations, is suggested to be aversive and the authors postulate that the repetitive behaviours characteristic of ASD may be understood as attempts to create rhythmicity in its absence. Empirical evidence of a positive effect of melatonin on stereotyped behaviours and behavioural rigidity (Garstang & Wallis, 2006; Malow et al., 2012) is considered to support this theory. The authors call for additional investigation of physiological rhythms in ASD and the examination of the implications of dysregulation in these rhythms for persons with autism.

Empirical Investigations of the Relation between Physiological Activity and Challenging Behaviour

The existence of such theories has prompted researchers to empirically investigate the relationship between physiological activity and challenging behaviour among persons with ASD. Of these studies, few have employed measures of LHPA axis activity. Verhoeven et al. (1999) assessed the association between LHPA axis functioning and the stereotyped and self-injurious behaviours of persons with intellectual disabilities. A trend towards lower cortisol levels among those exhibiting high rates of SIB or stereotypy was observed. Following this, Symons and colleagues (2003; 2011) compared the cortisol levels of individuals with developmental disabilities who engaged in severe SIB and controls who were diagnosed with developmental disabilities but who did not engage in

SIB. In both studies, higher cortisol levels were observed among those engaging in SIB. In their 2003 study, Symons et al. also reported a positive correlation between SIB and morning and evening cortisol levels.

Hall, Lightbody, and Reiss (2008) examined the relationship between cortisol levels, SIB, autism symptomatology, as measured by the Autism Diagnostic Observation Schedule (ADOS) which includes a measure of repetitive and restricted behaviours, and compulsive behaviour among 60 children diagnosed with Fragile X Syndrome. Neither SIB nor compulsive behaviours were correlated with cortisol levels.

Gabriels and colleagues (2013) investigated whether repetitive behaviours could account for the variability in cortisol levels observed among individuals with ASD in previous research studies (e.g. Corbett, Mendoza, Adullah, Wegelin, & Levine, 2006; Corbett, Schupp, Levine, & Mendoza, 2009; Kidd et al., 2012). Twenty-one children with ASD who presented with either high or low levels of repetitive behaviours participated. It was found that the overall cortisol levels of the two groups differed significantly such that children who presented with high levels of repetitive behaviours had lower cortisol levels, by 36%, than children who presented with low levels of repetitive behaviours. Neither age nor IQ had a significant effect on cortisol levels. The authors interpret these findings as support for a stress-reducing, calming function of stereotyped behaviour.

A greater number of studies have employed measures of ANS activity while investigating the link between physiological activity and challenging behaviours. Sroufe et al. (1973) appear to have conducted the first psychophysiological assessment of challenging behaviour. Their study examined the relationship between stereotypy, in the form of finger flicking, HR, behavioural indices of stress, such as muscular tension and facial expression, and environmental conditions, for a six-year-old boy diagnosed with ASD. It was found that stereotypy was consistently preceded by a brief HR deceleration and followed by a significant increase in HR, up to 32 beats per minute (bpm). HR

increases were greatest when stereotypy occurred during periods of rest or low physiological arousal which suggests that the HR patterns observed cannot be attributed to the effects of movement. The authors also proposed that the increased frequencies of stereotypy observed during new situations and during conflict suggest that stereotypy is related to stress which is supported by observations of accompanying muscular and facial tension during engagement in the behaviours. These results suggest that, for this participant, stereotypy served multiple functions including the elevation of physiological arousal during periods of low arousal and the release of excess energy or the blocking of additional environmental stimulation during periods of high arousal. In this study, psychophysiological measurement appeared to demonstrate an association between physiological arousal and stereotypy, contributed to the understanding of stereotypy's functional significance, and aided with treatment development and behaviour reduction.

Hutt, Forrest, and Richer (1975) investigated the relationship between HR and behaviour among nine children diagnosed with ASD. When compared to typically developing children, children with ASD were found to have higher resting HR. HR variability was also greater among participants diagnosed with ASD although it differed according to ongoing behaviour, being highest during stereotypy and lowest during task performance. The authors note that overt behaviour was most similar to that of typically developing participants, including less stereotypy and avoidance of social contact, when HR variability was most like that of typically developing participants. The authors interpret their results as being suggestive of a malfunctioning, sympathetically-dominated ANS that results in high levels of physiological arousal among individuals with ASD. This theory is further supported by the study's examination of HR patterns surrounding stereotypy which revealed that stereotypy typically resulted in HR decreases of five bpm or more. In this case, the repetitive movements involved in stereotypy appeared to function to reduce high levels of physiological arousal and to block further environmental stimulation.

Young and Clements (1979) recorded the HR, HRV, and stereotypy, in the form of complex hand movements, of three adolescents with severe intellectual disabilities during three conditions: 1) a restricted environment with minimal stimulation; 2) an enriched environment with a variety of play materials and an adult present, and 3) a treatment condition during which stereotypy resulted in the implementation of functional movement training, a brief form of overcorrection. The authors sought to determine, using psychophysiological measurement, whether stereotyped behaviours were "self-stimulatory" or "de-arousing". Stereotypy was most frequent in the restricted environment. Participants showed significantly decreased HR variability during complex hand movements and significantly increased HR variability following stereotypy. Both complex hand movements and body rocking, another form of stereotypy, were associated with increased HR during their occurrence. However, in contrast to complex hand movements, HR variability increased during body rocking. The authors therefore caution against assuming that all topographies of a class of behaviours are associated with the same physiological states. The authors conclude that the data support the theory that stereotypy serves a self-stimulatory function rather than functioning to reduce arousal but suggest that the function of the behaviour may differ according to diagnosis, such that individuals with intellectual disabilities are more likely to present with self-stimulatory stereotypy, while individuals with ASD are more likely to emit de-arousing stereotypy.

Lewis et al. (1989) researched the relationship between stereotypy, in the form of body rocking, and physiological activity in 17 individuals diagnosed with either severe or profound intellectual disabilities. HRV, but not HR, was significantly positively correlated with body-rocking rate. Body rocking did lead to increases in HR and HRV although the authors note that, considering its intensity and the gross motor movements involved, these were slight. The authors propose that the results of this study support the cardiac-somatic coupling hypothesis (Obrist, Webb, Sutterer, & Howard, 1970) which proposed that

individuals experiencing hypo-arousal may engage in motor movements to support efficient metabolic functioning.

Willemsen-Swinkels and colleagues (1998) examined HR patterns associated with stereotypy among 18 children diagnosed with pervasive developmental disorder, attention deficit disorder, ADHD, or a language disorder. They categorised the mood state associated with each occurrence of stereotypy as either “distress”, “elation” or “composure”. HR patterns associated with stereotypy differed depending on participant mood state during the behaviour. Stereotypy associated with distress tended to occur during states of high physiological arousal and to result in HR decreases. The authors suggest such stereotypy might be a response to excessive stimulation and might serve a calming function by reducing physiological arousal and diverting attention from an apparently stressful situation. Stereotypy associated with elation appeared to be preceded and followed by HR increases. The authors suggest that such behaviours might be understood as an expression of happiness or excitement. Stereotypy during periods of composure was not associated with HR changes and the authors suggest that this form of stereotypy might function to counteract under-stimulation or might have social functions.

Romanczyk and Matthews (1998) recounted several case studies of assessments that incorporated the psychophysiological measurement of challenging behaviour. They measured the EDA of two young girls, one diagnosed with autism, the other diagnosed with an intellectual disability and schizophrenia, who engaged in severe SIB. For one participant, high levels of physiological arousal were observed to consistently precede SIB. However, an undiagnosed ear condition was identified which, when treated, led to reductions in SIB. For the second participant, increases in physiological arousal in response to both physical contact and to periods of non-interaction led the researchers to hypothesise that the participant had difficulty in modulating physiological arousal. An intervention, incorporating continuous physiological measurement and real-time feedback

for staff, systematic desensitisation to the physical proximity of, or social interaction with, others and the creation of a quiet space to which the child could retreat when overwhelmed, led to near-zero rates of SIB and increases in appropriate social behaviours. Further psychophysiological assessments of two young children with ASD who presented with severe challenging behaviour which occurred in the absence of environmental antecedents or consequents were also conducted. However, the acquired data were of little use in treatment development as it was not possible to discern consistent physiological patterns that co-occurred with the behaviours. However, the authors note that problems with data collection, including participant noncompliance, using the physiological measurement devices were primarily responsible for the poor outcomes of these assessments.

Freeman et al. (1999) conducted a naturalistic study that examined the associations between challenging behaviours, such as SIB, aggression, and disruptive behaviours, HR, and environmental events for two males diagnosed with severe intellectual disabilities. Occurrences of challenging behaviour were consistently followed by HR increases while it was not possible to identify a specific HR pattern which consistently preceded challenging behaviour. A significant negative correlation between mean HR and frequency of challenging behaviours was also observed for one participant such that challenging behaviour was more frequent when HR was lower than when HR was higher. Higher HR was typically associated with overt frustration and distress while lower HR was associated with overt happiness, energy, and excitement. The authors propose that challenging behaviour may act as a discriminative stimulus for increases in physiological arousal. The results do not support suggestions that an increase in HR, or physiological arousal, precedes challenging behaviour and serves as an antecedent for challenging behaviour. However, Freeman et al.'s (1999) analyses were potentially limited as their HR monitor only recorded HR at 15 second intervals which may not have been sensitive enough to

capture the HR changes surrounding challenging behaviours. Freeman and colleagues (2001) conducted a more detailed analysis of additional data from one of the participants from their prior study and found that high HR during the behaviour or in the preceding 30 seconds predicted the occurrence of SIB. They also identified an interaction between environmental and physiological influences such that SIB in response to external demand was more likely if HR was high. The data also indicated that challenging behaviour was unlikely to occur, even when HR was high, if there were no ongoing external activities. This analysis suggested that physiological arousal may be an antecedent variable which provokes SIB or that it may act as a setting event which increases the aversiveness of external stimuli, such as demand, and makes engagement in the challenging behaviour negatively reinforcing.

Hirstein and colleagues (2001) examined the EDA of 37 children with ASD as they sat quietly, interacted with their parents, immersed their hands in a bowl of dry beans, and engaged in their preferred self-stimulatory activities such as watching a favourite video, arranging books or toys, or flicking through a book. The authors identified two distinctly different types of responders among their sample. The first group, comprised of 26 of the 37 participants, presented with elevated EDA, which was highly variable, and large skin conductance responses. Interactions with others led to increased EDA while behaviours such as the immersion of hands in a bowl of beans, eating, sucking on sweets, being wrapped in a blanket, and deep pressure massage, led to reductions in EDA. Interruptions to preferred activities typically led to extremely large skin conductance responses that were often followed by challenging behaviour. In the second group, four of the 37 participants, had extremely low EDA and little electrodermal responsivity to the various activities. Among these participants, only intense activities such as SIB led to changes in EDA. The authors propose that, among children with ASD, differentially responsive subgroups exist such that some children may be physiologically hyper-aroused and hyper-reactive and

other children may be physiologically hypo-aroused and hypo-reactive. For the hyper-aroused group, self-stimulatory activities appeared to be calming and to reduce physiological arousal. Their large physiological responses to interruptions, or changes, suggest that the co-occurring challenging behaviour and distress may be the result of dramatic, uncomfortable, increases in physiological activity. For the hypo-aroused group, SIB and other intense behaviours appear to function to increase physiological activity. The authors propose that stereotypy, SIB, and other challenging behaviours may serve to regulate a malfunctioning ANS. Some children with ASD may seek out repetitive activities or engage in stereotypy in order to calm an over-active ANS while others may engage in high-intensity behaviours such as SIB in order to activate a lethargic ANS.

Barrera and colleagues (2007) recorded HR, every 1 second, during functional analyses of the severe SIB of three adults with developmental disabilities, including ASD, pervasive developmental disorder, and cornelia de lange syndrome, and severe or profound intellectual disabilities. An identical HR pattern was associated with SIB for all three participants although it did vary in strength. This pattern consisted of HR increases and peak prior to, or during SIB, and a significant decrease in HR following SIB. Detailed analyses revealed that this HR pattern was not attributable to movement, HR level, respiratory activity, stance, the topography of the SIB, or the duration of SIB. External operant contingencies during the functional analyses had little effect on the observed HR waveform. The authors propose that elevated physiological arousal served as a precursor, or antecedent, for SIB which functions to decrease physiological arousal. In this way, SIB appeared to be maintained by negative reinforcement as engagement in the behaviour served to terminate aversive levels of elevated arousal, findings which support previous researchers' (Brain et al., 1998; Haines et al.,1995) suggestions that SIB serves to reduce internal tension.

Hoch and colleagues (2010) conducted an assessment of the relationship between physiological arousal, measured using HR, and activity choice for a young boy diagnosed with ASD. The authors found that the participant was more likely to choose highly arousing activities when he was highly aroused and more likely to choose minimally arousing activities when he was in a low state of arousal. Of relevance to the proposed research, SIB was more probable when the participant was highly aroused than when he was less aroused.

Jennett, Hagopian, and Beaulieu (2011) monitored the HR, recorded every 30 seconds, of a young girl diagnosed with ASD during SIB. They reported that the participant exhibited overt signs of anxiety during SIB and hypothesised that SIB may be associated with increased levels of physiological arousal. The authors assessed whether physiological state during SIB differed depending on whether the participant was restrained or not. They found that when restraint was applied, HR remained near resting levels and SIB did not occur. When restraint was removed, HR increased significantly, up to 30 bpm, along with corresponding increases in SIB. HR returned to resting levels when restraints were re-applied. The authors propose that the increased physiological arousal recorded when restraints were removed was an anxiety response due to the increased likelihood of SIB which appeared aversive to the participant. Thus, SIB may have been negatively reinforced by the application of restraints which reduced the likelihood of further SIB and led to HR decreases.

Lydon and colleagues (2013) conducted a naturalistic study which investigated the association between HR, measured every 5 seconds, and the challenging behaviours of three youths diagnosed with ASD and co-morbid intellectual disabilities. Challenging behaviours recorded included stereotypy, SIB, destructive behaviour, and self-management behaviours. As in Freeman et al.'s (1999) study, SIB instances were typically followed by HR increases. HR was elevated during SIB for two of the three participants which may be

attributable to stress caused by the antecedent stimuli or the pain caused by the SIB. Stereotypy was typically preceded and followed by HR increases for two of the participants, while HR increases were only observed post-stereotypy for the third participant. HR during stereotypy was higher than mean HR for all three participants. The authors note that the findings offer little support for homeostatic theories of stereotypy and instead propose that these findings may indicate that some individuals diagnosed with ASD may enjoy the feelings and bodily sensations that accompany high physiological arousal and that behaviours such as stereotypy and SIB may be positively reinforced by the increases in physiological arousal which they engender. A subsequent study by Hall and colleagues (2013) produced similar findings. During a functional analysis which incorporated HR measurement, the authors observed that the SIB in the form of skin picking, of a young male with prader-willi syndrome was positively reinforced by the increases in physiological arousal which it produced.

Moskowitz and colleagues (2013) examined the relationship between overt signs of anxiety, challenging behaviour, and HR measures. Three male children, 6-9 years of age, diagnosed with ASD, a co-morbid intellectual disability, and meeting the criteria for the diagnosis of an anxiety disorder participated. Topographies of challenging behaviour measured during the analysis included yelling or screaming, elopement, aggression, tantrum behaviour, and inappropriate behaviours in the car. Overt indicators of anxiety included clinging to the caregiver, crying or tearfulness, cowering, repetitive questioning, verbal pleading, plugging of the ears, eyes rapidly darting back and forth, and finger mouthing. During the analysis, participants were exposed to conditions that were established as being either “high-anxiety” or “low anxiety” situations. For all participants, HR was significantly higher during the “high anxiety” condition than during its “low anxiety” counterpart. Further, challenging behaviours occurred at much higher rates during the high anxiety condition than during the low anxiety condition. The authors highlight the

importance of context in the assessment of behavioural function; in this case, certain challenging behaviours were indicative of anxiety in certain contexts but not in others. The authors further suggest that the results of this analysis indicate that anxiety may act as an S^d (discriminative stimulus) for challenging behaviour or a setting event.

Finally, Hoch et al. (2013) examined the relationship between HR, HRV and SIB among two children diagnosed with ASD and a co-morbid intellectual disability and an additional child diagnosed with a primary diagnosis of an intellectual disability. Indices of HR were correlated with SIB for two of the participants. However, HR was found to be related to SIB for only one of the participants, a 9 year old boy diagnosed with autism, a co-morbid intellectual disability, and cerebral palsy. For this participant, SIB was most likely to follow a HR increase. This pattern was significantly more likely to occur than its alternatives. An assessment of the relationship between general motor movement and HR did not suggest that it could have accounted for this relationship.

Conclusions and Research Aims

While applied behaviour analysis (ABA) has yielded successful treatments for the challenging behaviour of persons diagnosed with autism, the aetiology of such behaviours remains primarily theoretical with little behavioural research directed at determining the factors responsible for the emergence of problem behaviour. This criticism of applied behaviour analytic research is not new; the need for a better understanding of challenging behaviours, such as stereotypy and SIB, and their development, and the inadequacy of a solely behavioural account of these disorders have been highlighted by researchers in the field over the past decades (Cataldo & Harris, 1982; Devine & Symons, 2013; Duerden et al., 2012; Favell et al., 1982; King, 2000; MacLean et al., 1994; Rapp and Vollmer, 2005b; Schroeder & Tessell, 1994). Barrera and colleagues (2007) emphasised that ABA needs to rethink its current treatment-orientation and to devote more attention to assessment, such as that of the relationship between biological variables and challenging behaviour, so as to

further the field's understanding of the aetiology of behavioural problems so frequently encountered and treated. The current body of research suggests that the examination of physiological activity may yield a better understanding of the challenging behaviours of at least some persons diagnosed with autism. The current research program may also be considered timely as low-arousal interventions for challenging behaviour, predicated upon the notion than high levels of physiological arousal contribute to engagement in challenging behaviours, have become popular in recent years although the relationship between physiological arousal and challenging behaviour requires further empirical investigation (see McDonnell, McCreadie, Mills, Deveau, Anker, & Hayden, 2015 for a review of the Low Arousal Approach to the treatment of challenging behaviour).

The proposed research thus aims to further examine the relationship between physiological activity and challenging behaviour among children and adolescents diagnosed with ASD and is centred around the question "what is the explanatory value of physiological data, and what new information can the data provide?", posed by Romanczyk and Matthews (1998, p.117). The aims of the present research program are: (1) to conduct a systematic review of PR to stimuli among persons with autism and its potential implications for our understanding of associated atypical and challenging behaviours; (2) to examine the relationship between measures of ANS and LHPA axis activity and to assess whether levels of, or changes in, physiological activity may act as antecedents to or reinforcers for challenging behaviours, and (3) to examine the correspondence between overt behaviour, mood state, and physiological activity. These research studies are also designed to remedy the deficits of previous research studies in this area; Cohen and colleagues (2011) have previously suggested several methodological improvements including the use of single-subject analyses, assessment under naturalistic conditions, reinforcement for compliance with assessment procedures, the use of multiple measures of ANS activity, the measurement of overt affective state alongside physiological

measurement, and the documentation of factors, such as medical conditions, medication use, co-morbid psychological disorders etc., which may influence physiological responses.

Chapter 2: Study 1

A Systematic Review of Physiological Reactivity to Stimuli in Autism Spectrum Disorder[1]

[1] This chapter has been accepted for publication: Lydon, S., Healy, O., Reed, P., Mulhern, T., Hughes, B. M., & Goodwin, M. S. (in press). A systematic review of physiological reactivity to stimuli in autism. *Developmental Neurorehabilitation.* doi: 10.3109/17518423.2014.971975

Abstract

The prevalence of abnormal behavioural responses to a variety of stimuli among individuals with autism has led researchers to examine whether physiological reactivity is atypical in this population. The current systematic review examines published literature assessing physiological reactivity to sensory, social and emotional, and stressor stimuli in individuals with autism. A novel measure of methodological quality suitable for use with non-randomised, non-interventional, psychophysiological studies was also developed and applied to the reviewed studies. Across 57 studies that met our inclusion criteria, individuals with autism were found to respond differently than typical developing populations in 78.6%, 66.7%, and 71.4% of these stimulus classes, respectively. However, this extant literature is characterised by variable and inconsistent findings, which do not appear to be accounted for by varying methodological quality, making it difficult to determine what specific factors differentiate individuals with autism who present with atypical physiological reactivity from those who do not. Despite this uncertainty, individual differences in physiological reactivity are clearly present in autism, suggesting additional research is needed to determine the variables relating to physiological activity among those with ASD and to examine the possible existence of physiological subtype responders in the population.

Behavioural hypo-reactivity (e.g., lack of reaction to human speech, loud noises, pain) and hyper-reactivity (e.g., heightened sensitivity, agitation or distress in response to particular clothing or food textures, or to everyday noises) to stimuli have long been described in individuals with autism (Liss, Saulnier, Fein, & Kinsbourne, 2006). Abnormal behavioural reactions are considered characteristic of autism and oftentimes used as criteria that distinguish the condition from other developmental disorders, such as intellectual disability (Klintwall et al., 2011; Wiggins, Robins, Bakeman, & Adamson, 2009). Furthermore, abnormal behavioural reactivity has been linked to several negative outcomes in autism, including internalising and externalising behaviours that complicate participation in typical childhood leisure, social, and educational activities (Reynolds, Bendixen, Lawrence, & Lane, 2011; Tseng, Fu, Cermak, Lu, & Shieh, 2011). Several researchers have hypothesised that abnormalities in PR may underlie behavioural issues in autism, wherein hyper-arousal is associated with experiences of fear, anxiety, and avoidance (Dalton et al., 2005; Hutt & Hutt, 1965; Hutt & Ounsted, 1966; Hutt et al., 1964) and hypo-arousal is associated with feelings of dullness, under-stimulation, and sensory seeking (Rogers & Ozonoff, 2005; Rimland, 1964). Moreover, researchers have postulated that challenging behaviours such as aggression, self-injury, tantrums, elopement, and stereotypy are associated with PR (for a review see: Cohen et al., 2011). Taken together, the existence of and diversity across these theories, increasing prevalence of autism, and behavioural problems associated with the condition all suggest a need for further examination of PR in this population, and serves as the impetus for the present review.

PR has been defined as “the deviation of a physiologic response parameter from a comparison or control value that results from an individual’s response to a discrete, environmental stimulus” (Matthews, 1986, p.114). In this way, PR may be conceptualised as the difference between physiological activation in response to a stimulus or stressor as

compared to resting or baseline levels. PR is most commonly measured by assessing ANS and LHPA axis activation. The ANS comprises sympathetic, parasympathetic, and enteric branches. To our knowledge little is known about the enteric system in autism, therefore we include no further discussion of it in this review. The SNS is responsible for activation and mobilisation of the body to facilitate attention, fight, and flight. The PNS is responsible for recovery and restoration. The LHPA axis regulates bodily responses to stress and promotes restoration of homeostasis following a stressor. The role of the LHPA axis can be considered protective (Manuck & Krantz, 1986); it ensures that the body returns to normal physiological functioning following a stressor to prevent damage to the body from its own physiological reaction to stress. Changes occur quickly in the ANS in response to stimulation, while the LHPA axis response is significantly slower. Typically, the two systems are thought to co-activate in response to stress or stimulation. However, while correlations between LHPA axis and ANS responses have been recorded (Pasquali et al., 1996), other studies find little association between them (van Goozen et al., 1998).

Activation of the ANS in response to stimulation or stress is most commonly measured through HR, HRV, BP, and EDA, while LHPA system functioning is typically assessed through measurement of cortisol (Boyce & Ellis, 2005; Lovallo, 2005; Ordaz & Luna, 2012). Although PR is highly individually variable, all of these measures have at least some degree of temporal stability (Alkon et al., 2003; Burleson et al., 2003; Schell, Dawson, Nuechterlein, Subotnik, & Ventura, 2002). Furthermore, a number of specific ANS and LHPA axis response patterns have been found to associate with an individual's perception of, response to, and putative feelings about stimuli in the environment. Slowing of HR and a corresponding increase in EDA is associated with attention and interest (Eisenberg & Fabes, 1990; Fabes, Eisenberg, & Eisenbud, 1993; Lang, Greenwald, Bradley, & Hamm, 1993). Defensive reactivity, in the form of adaptive physiological mobilisation allowing an individual to respond behaviourally, indicates the perception of a

stimulus as threatening or harmful (Lang, Bradley, & Cuthbert, 1997). Perceptions and feelings of stress and anxiety are manifested through SNS and LHPA axis activation as reflected through increased HR, BP, EDA, and production of cortisol, and concurrent decreases in HRV. Habituation is associated with attenuation of hormonal and physiological responses following repeated presentation of a stimulus or set of stimuli (Cyr & Romero, 2009). Habituation to stimuli perceived to be stressful is highly important, as long-term physiological activation in response to stress has been linked to changes in behaviour, disruption of normal physiological functioning, and disease (Cyr & Romero, 2009; McEwen, 1998).

PR has been shown to play an important role in a variety of areas relating to typical child development. For instance, Kagan and colleagues (1987) identified a relationship between patterns of social interaction among children and their internal physiological activation such that behaviourally inhibited children, who were shy and fearful, could be differentiated from behaviourally uninhibited children, who were extraverted and fearless, on the basis of their respective LHPA axis or SNS activation. Fox (1989) demonstrated that PR was associated with emotional reactivity and sociability among infants such that greater PR to positive or negative events at five months was associated with greater sociability at 14 months. Hart and colleagues (1995) found that cortisol reactivity in maltreated and socially deprived children was positively correlated with social competence and negatively correlated with shyness and internalising behaviour. Keenan and colleagues (2009) have proposed that poor or faulty modulation of PR to stimuli in the early years may be a risk factor for later development of behavioural or emotional problems, with empirical findings suggesting that cortisol reactivity is related to maladaptive behaviour in typically developing infants.

Studies have also identified PR as a risk factor for a variety of atypical behaviours and psychiatric comorbidities relevant to autism. Bauer and colleagues (2002) reviewed

studies examining correlations between PR and behaviour and concluded that low levels of PR and baseline arousal are associated with externalising symptoms, such as aggression or disruptive behaviours, while high levels of PR and baseline arousal are associated with internalising symptoms, such as social withdrawal and anxiety. A meta-analysis by Lorber (2004) concluded that high EDA reactivity, low baseline HR, and high HR reactivity are associated with aggression and conduct problems. Other research has identified associations between PR and a variety of psychiatric disorders including anxiety, oppositional defiant disorder, depression, and post-traumatic stress disorder (Monk et al., 2001; Sapolsky, 2000; van Goozen et al., 1998). High rates of co-morbid psychiatric disorders are commonly reported in individuals with autism (Simonoff et al., 2008) suggesting that PR may contribute to their development, emergence, and/or expression.

Given the developmental, behavioural, and psychiatric significance of PR, the purpose of the current paper was to systematically review, quantitatively synthesise, and evaluate the methodological quality of extant research focusing on cardiovascular, electrodermal, and hormonal measures of PR in individuals with autism in response to different types of stimulus presentations. We conclude with open questions, research challenges to be overcome, and suggestions for future research.

Method

Search Procedures

We identified articles by conducting comprehensive searches of PsycInfo, Psychology and Behavioural Sciences Collection, Medline, Scopus, and Web of Science. Searches were carried out by inputting *autism* in combination with the following key words: *stimuli*, *habituation*, *orientation*, *reactivity*, *responsivity*, *physiology*, *autonomic*, *psychophysiology*, *stress*, *arousal*, *hyperarousal*, *hypoarousal*, *heart*, *cardiac*, *blood pressure*, *skin conductance*, *galvanic skin response*, *electrodermal*, and *cortisol*.

Publication year was not restricted but only papers published in the English language were considered for inclusion.

Inclusion and Exclusion Criteria. Searches were limited to peer-reviewed journals. We included studies if they: (1) had at least one participant diagnosed with autism; (2) measured either HR, HRV, respiratory sinus arrhythmia, BP, EDA, or cortisol; and (3) used research designs that exposed participant(s) to at least one stimulus condition different from baseline. We excluded studies if they: (1) only measured baseline physiological activity; (2) were described as a case study; (3) investigated physiological activity during challenging behaviour; (4) utilised physiological measures to assess the effects of a treatment or pharmacological intervention; (5) grouped participants with autism with non-autistic participants for analysis; (6) compared the PR of those with autism to a control group comprised of individuals with other psychological diagnoses (e.g., intellectual disabilities, developmental delays) only, and (7) examined PR to more than one type of stimulus and did not present findings to each stimulus type separately. In studies that utilised a typically developing control group along with one or multiple control groups with other diagnoses, only data on the autism and typically developing groups were extracted for the purposes of this review.

The search procedures produced 128 articles that utilised physiological measures with persons diagnosed with autism. After filtering them using the process presented in Figure 1 which depicts the inclusion/exclusion criteria employed, 57 studies remained and were subsequently sorted into the following three stimulus classes: *Sensory stimuli* were defined as any stimulus perceived by an individual's senses such as pictures, lights, sounds, tastes, movements, odors, or textures. *Social and emotional stimuli* were defined as a stimulus involving other individuals, elements of social interaction (e.g., eye gaze or speech), or human affect or emotion. *Stressor stimuli* were defined as any situation hypothesised or intended to cause participants stress. The accuracy of the data extracted

was determined by having a second rater review all studies and calculating interrater reliability. Interrater reliability was found to be 94.73% (range 74.1%-100%). In cases of non-agreement, consensus was achieved between raters through review and discussion.

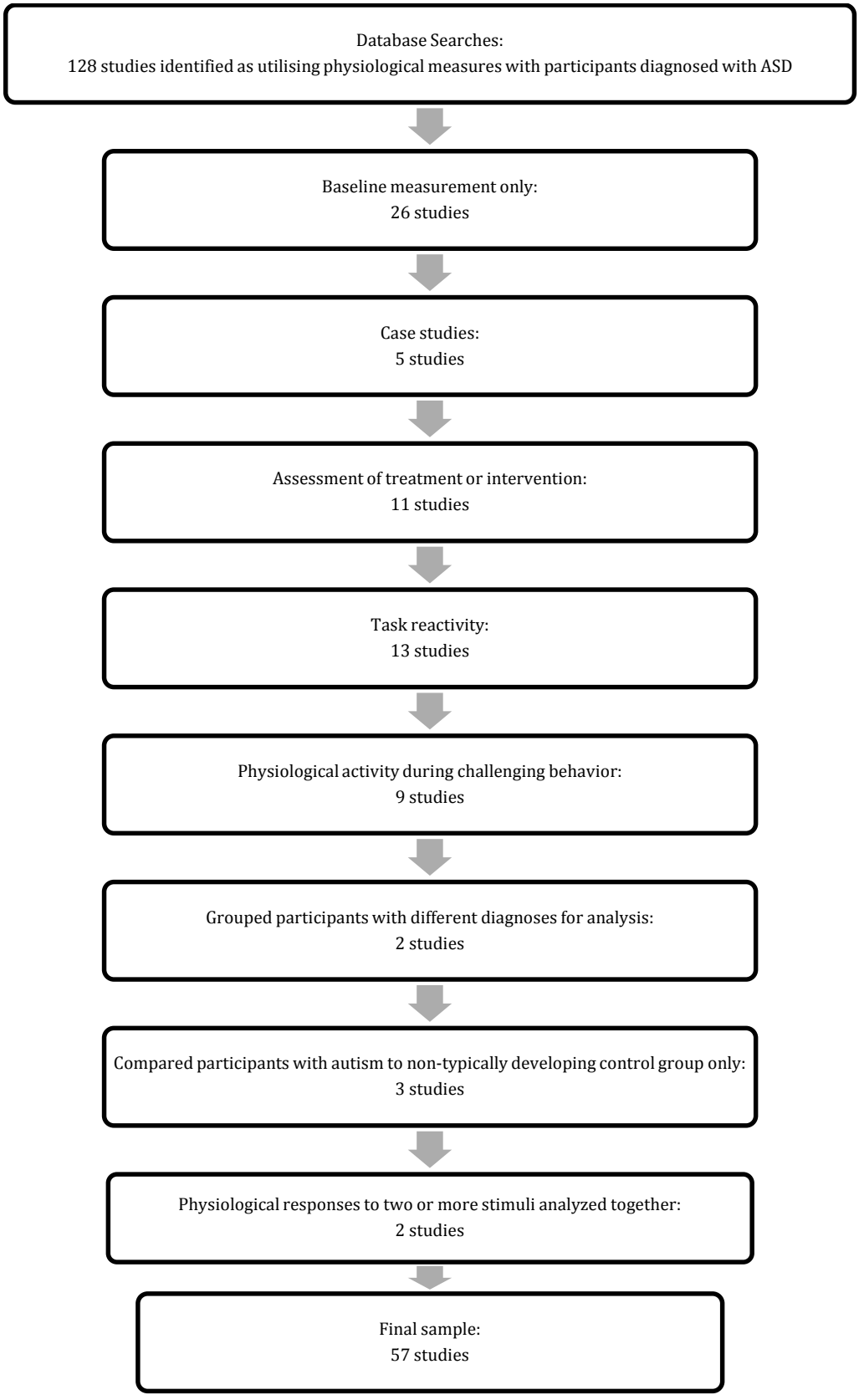

Figure 1. Flow diagram showing inclusion/exclusion of studies identified during database search process.

Quality Assessment

The importance of assessing the methodological quality of studies included in systematic reviews is widely recognised and plays a key role in the derivation of conclusions from existent research (Farrington, 2003; Higgins, 2008; Moher, Liberati, Tetzlaff, & Altman, 2009). Extensive literature searches conducted during the preparation of the manuscript failed to identify any existing quality assessment instrument suitable for the application to non-randomised, non-interventional, psychophysiological research studies such as those included in the current review. For this reason, a quality assessment measure suitable for use with the included studies was devised in accordance with Farrington's (2003) suggestions for the assessment of methodological quality standards and the deficits or faults of research in this area which have been previously identified by Cohen and colleagues (2011). Table 1. presents the quality assessment measure which was applied to the included studies. It is comprised of 22 items, separated into four of the key categories of methodological quality identified by Farrington (2003). Three of the items are comprised of multiple related sub-items. In total, raters were required to make 33 judgments or evaluations for each study, marking each criterion as present or absent with the exception of the external validity criterion which was scored as either 3 (very good), 2 (adequate), 1 (poor), or 0 (very poor). The measure was scored by calculating the number of items that were met per category. If 91% or more of the criteria in a category were met then the study received a rating of three for that category indicating a "very good" outcome. A rating of two was assigned to studies that met 71-90% of the criteria indicating an "adequate" outcome on that category. A rating of one was assigned to studies which met 51-70% of the criteria in a category indicating a "poor" outcome on that category, and a rating of zero indicated studies which met 50% or less of the criteria within a category resulting in a "very poor" outcome on that category. In this way, the highest possible quality rating achievable was 12 points. Within each stimulus category, a median split was

performed subsequent to scoring whereby studies receiving a score equal to, or higher than, the median were classified as being of higher quality and studies receiving a rating below the median score were classified as being of lower quality. Interrater reliability was calculated to assess the agreement across all criteria of the quality assessment, for all included studies, between two independent raters. The mean interrater agreement for each study was 94.3% (range 84.8-100%). In cases of non-agreement, consensus was achieved between raters through discussion. The quality assessment provided a differentiation of studies within each of the three stimulus classes according to high and low methodological quality. The analysis of studies identified as being of high methodological quality within each stimulus class, allowed us to examine whether methodological rigor impacted upon experimental outcomes, and to provide more robust conclusions on the existence of abnormalities in PR among those with ASD compared to typical developing controls.

Table 1

Criteria used to Assess the Methodological Quality of the Included Studies

	Criteria
Descriptive Validity	➢ Experimental design is stated ➢ Sample size is stated ➢ The following participant characteristics are outlined: a) Age b) Gender c) Co-morbid medical and psychological diagnoses d) ASD severity e) Intellectual functioning f) Medication use ➢ Background factors which affect physiological responses are measured: a) General physical fitness b) Affective state during the experimental procedures c) Physical activity during the experimental procedures ➢ The physiological response, and any behavioural responses being measured, are operationally defined ➢ The stimulus/stimuli presented are described in detail including information on their intensity and duration ➢ If standardised measures are used, the psychometric properties of these are described ➢ Statistical methods employed are outlined
Internal Validity	➢ A control group or control condition is utilised ➢ If a control group is used, an attempt to match the experimental and control group on pertinent variables is made ➢ Consideration of potential mediator or moderator variables within analyses is evident ➢ Background factors which impact physiological responses are controlled for either through procedural consideration or analyses: a) Psychological or medical conditions b) Medication use c) General physical fitness d) Affective state e) Physical activity during experimental session ➢ Baseline physiological activity is considered during analyses ➢ Experimental procedures are conducted in a natural or familiar setting or an effort to habituate participants to the experimental setting prior to data collection is documented ➢ An attempt is made to habituate participants to the physiological recording device prior to data collection ➢ Multiple measures of physiological activity are employed during experimental procedures
External Validity	➢ Experimental stimuli are representative of those which may be encountered in everyday life
Statistical Conclusion Validity	➢ The statistical significance of findings is examined ➢ Effect sizes are calculated for findings ➢ Confidence intervals are calculated for findings ➢ Analyses appropriate for the research question are utilised ➢ If group analyses are employed, individual responding is also examined.

Results

Sample Characteristics

Sample sizes ranged from 3-188 participants with a mean of 45.3 participants per study among those studies reporting total sample size. A weighted mean age was calculated for the total sample of each study. We found 33 studies (57.9%) included children (1- 11 years), 11 studies (19.3%) included adolescents (12-17 years), and eight studies (14%) included adults (18+ years). Insufficient data were reported in five studies (8.8%) to calculate weighted mean age of all participants. Participants with autism as a primary diagnosis were included in all of the studies. Participants with asperger syndrome were included in 10 studies (17.5%). Participants with PDD-NOS were included in nine studies (15.8%). Five studies (8.8%) did not employ a control group, while the remaining 52 studies (91.2%) employed typically developing control groups.

Physiological Measures

The majority of studies (59.6%) used only one physiological measure. EDA was used in 52.6% of studies, HR in 40.4% of studies, and cortisol in 24.6% of studies. Measures of respiratory sinus arrhythmia, HRV, and BP were less frequent, being employed in 12.3%, 5.3% and 1.8% of studies respectively. Other physiological measures employed were respiration, heart period, pre-ejection period, interbeat interval, peripheral vasoconstriction, cephalic vasodilation, peripheral blood flow, vagal tone, evoked cardiac response, evoked cardiac deceleration, urinary mucoprotein excretion, temperature, neuroendocrine measures, EEG, and plasma β-endorphin. The minority of studies (40.4%) employed more than one physiological measure. Combined use of HR and EDA was observed in seven studies (12.3%) and HR and cortisol was observed in four studies (7%). Participants with autism showed responses statistically significantly different from controls in 65.5% of studies employing EDA; 50% of studies employing HR; and in 53.8% of

studies employing cortisol. All of these primary physiological measures thus seemed similarly sensitive to PR differences in participants with autism.

Sensory Stimuli

Fifteen studies assessing PR to sensory stimuli are presented in Table 2. Participants with autism could be distinguished from controls in 11 of 14 controlled studies (78.6%) (Barry & James, 1988; Chang et al., 2012; James & Barry, 1984; Palkovitz & Wiesenfeld, 1980; Schaaf, Benevides, Leiby, & Sendecki, 2015; Schoen, Miller, Brett-Green, & Nielsen, 2009; Stevens & Gruzelier, 1984; van Engeland, 1984; van Engeland, Roelofs, Verbaten, & Slangen, 1991; Woodard et al., 2012; Zahn, Rumsey, & van Kammen, 2012). Across physiological measures employed, participants with autism were similar to controls in EDA, HR, and HRV (38.9%) when exposed to auditory stimuli, odorous stimuli, and the sensory challenge protocol (Palkovitz & Wiesenfeld, 1980; Zahn et al., 1987; Allen, Davis, & Hill, 2013; Legisa, Messinger, Kermol, & Marlier, 2013; McCormick et al., 2014); showed increased PR in EDA and HR (22.2%) when exposed to visual, auditory, olfactory, kinesthetic, tactile, and gustatory stimuli (Barry & James, 1988; Chang et al., 2012; James & Barry, 1984; Woodard et al., 2012); decreased PR in EDA (11.1%) when exposed to visual stimuli and the sensory challenge protocol (Schoen et al., 2009; van Engeland et al., 1991); and different patterns of PR among participants with autism in HR and EDA (27.8%) when exposed to various auditory stimuli and the sensory challenge protocol (Palkovitz & Wiesenfeld, 1980; Schaaf et al., 2015; Stevens & Gruzelier, 1984; van Engeland, 1984; Zahn et al., 1987). With respect to participants with autism only, one study demonstrated the absence of a physiological orienting response when exposed to auditory stimuli of varying social importance (Palkovitz & Wiesenfeld, 1980); two studies found delays in stimulus registration when exposed to auditory stimuli (Stevens & Gruzelier, 1984; van Engeland, 1984); three studies observed slow or absent habituation when exposed to auditory or visual stimuli (Barry & James, 1988; James &

Barry, 1984; Stevens & Gruzelier, 1984); two studies reported faster habituation when exposed to auditory stimuli (van Engeland, 1984; Zahn et al., 1987); one study reported less physiological changes from one stimulus to another among participants with ASD than among controls (Schaaf et al., 2015).

Table 2

Summary of Studies utilising Sensory Stimuli

Study	*n*	Age range (mean)	Experimental Group Diagnoses	Control Group	Stimuli	Physiological Measure(s)	Findings
Allen et al. (2013)	47	- (34.7 years)	High-functioning autism	Typically developing	Auditory (musical stimuli)	EDA	Similar EDR to the stimuli in both groups.
Barry and James (1988)	32*	4.9-17.2 years -	Autism	Typically developing	Visual and auditory stimuli	EDA; Respiration; Vasoconstrictive peripheral pulse amplitude response	Participants with autism demonstrated stronger PR to stimuli and did not show physiological habituation to the stimuli as control did.
Chang et al. (2012)	50	5-12 years (8.2 years)	High-functioning autism	Typically developing	Auditory stimuli	EDA	The autism group was significantly more aroused at baseline; The autism group demonstrated significantly stronger PR to auditory tones (excepting sirens) than the control group; Physiological activity was significantly higher during the recovery period for participants with ASD; Auditory under- and over-responsiveness (Sensory Processing Measure-Home Form) were both positively correlated with PR to the auditory stimuli.
James and Barry (1984)	80	4.5-16.9 years -	Autism	Typically developing	Visual and auditory stimuli	EDA; Respiratory period; Evoked cardiac response; Vasoconstrictive peripheral pulse amplitude response	PR to the stimuli was significantly stronger among the autism group than among controls. A failure to habituate to repeated stimulus presentations was also noted among participants with ASD.
Legiša et al. (2013)	16	8-14 years -	High-functioning autism	Typically developing	Odorous stimuli	HR; EDA	The groups did not differ on PR to the stimuli.

McCormick et al. (2014)	87	2.4-4.7 years (3.3 years)	Autism Spectrum disorder	Typically developing	Sensory challenge protocol	EDA	The groups did not differ on baseline physiological activity; The groups did not differ on PR to any of the stimuli. Habituation to repeated stimulus presentation was evident in both groups; There was no correlation between behavioural measures (Short Sensory Profile; Repetitive Behaviours Scale-Revised) and PR.
Palkovitz and Wiesenfeld (1980)	20	5.8-10 years (7.6 years)	Autism	Typically developing	Auditory stimuli of varying social importance	HR; EDA	Baseline EDA was significantly higher in the autism group while HR was similar in both groups; Control participants showed an orientation response in HR to all stimuli while children with autism did not. EDR to the stimuli was similar in both groups; Recovery of baseline HR was similar in both groups.
Schaaf et al. (2015)	88	6-9 years (7.8 years)	Autism; Asperger syndrome; PDD-NOS	Typically developing	Sensory Challenge Protocol	RSA; Pre-ejection period	The ASD group was significantly more aroused at baseline; The ASD group showed less change in RSA from stimulus to stimulus than controls did; The ASD group were observed to demonstrate behavioural dysregulation in response to the stimuli which corresponded with their lack of flexibility in physiological responding.
Schoen et al. (2009)	71	4-15 years (8.8 years)	High-functioning autism or Asperger Syndrome	Typically developing	Sensory challenge protocol	EDA	Baseline EDA was significantly lower in the autism group; The autism group showed consistently lower EDR to the sensory stimuli than the control group; There was no correlation between sensory behaviours and PR to stimuli (Short Sensory Profile).
Schoen et al. (2008)	38	5-15 years (9 years)	High-functioning autism or Asperger syndrome	-	Sensory challenge protocol	EDA	Results suggest the presence of hypo-aroused participants (low baseline EDA, weak PR to stimuli, slow to respond to stimuli and quick to habituate) and hyper-aroused participants (high baseline EDA, short latency to respond, strong PR to stimuli, and slow to habituate to stimuli).

Stevens and Gruzelier (1984)	20*	7-17 years (10.9 years)	Autism	Typically developing	Auditory	EDA	Participants with ASD appeared to show delays in stimulus registration by responding more strongly to the second tone than the first. They were also slower to habituate to stimuli than controls.
van Engeland (1984)	80	- (8.8 years)	Autism	Typically developing	Auditory	EDA	There were no differences in spontaneous electrodermal fluctuations pre-stimulation; The autism group showed delays in stimulus registration. The groups did not differ on habituation, EDR, or recovery. There were significantly more non-responders in the ASD group than in the control group.
van Engeland et al. (1991)	40	- (9.9 years)	Autism	Typically developing	Visual	EDA	The autism group showed significantly weaker EDR to stimuli. However, manipulation of stimulus subjective significance led to increased EDR. The groups did not differ on habituation to stimuli; No correlation between IQ and EDR was observed.
Woodard et al. (2012)	16	2-3.2 years (2.7 years)	Autism	Typically developing	Visual; Auditory; Olfactory; Kinesthetic ; Tactile; Gustatory	HR	Physiological measures indicated that children with autism were generally more hyper-reactive and less hypo-reactive than controls in response to sensory stimuli; There was a weak negative correlation between Low Registration (Infant/Toddler Sensory Profile) and PR.
Zahn et al. (1987)	32	18-39 years (27.6 years)	High-functioning Autism	Typically developing	Auditory Stimuli	EDA: HR; HRV; Respiration; Skin temperature	Groups were similarly aroused in EDA and HR at baseline but the autism group had higher HRV; Orientation and PR to the stimuli were similar among participants with autism and controls. However, participants with autism tended to habituate to the stimuli more quickly.

* information relates to autism group only, data on control group not provided. EDA = Electrodermal Activity, EDR = Electrodermal Reactivity, HR = Heart Rate, HRV = Heart Rate Variability, PR = Physiological Reactivity, RSA= Respiratory Sinus Arrhythmia

An examination of the 10 studies examining PR to sensory stimuli which were categorised as “high” quality (Allen et al., 2013; Barry & James, 1988; Legisa et al., 2013; McCormick et al., 2014; Schaaf et al., 2015; Schoen et al., 2008; Schoen et al., 2009; van Engeland et al., 1991; Woodard et al., 2012; Zahn et al., 1987) revealed that the exclusion of studies of lower quality led to a reduced proportion of studies (66.7% from 78.6%) suggesting abnormalities in PR among those with ASD as compared to typically developing controls. In addition, many of the studies suggesting issues with orientation, stimulus registration, or habituation were eliminated; only two remaining studies were suggestive of faulty habituation processes (Barry & James, 1988; Zahn et al., 1987), and one study suggestive of less physiological changes from one stimulus to another among participants with ASD (Schaaf et al., 2015). A non-controlled study by Schoen et al. (2008), which identified the presence of hypo- and hyper-aroused participants among their sample of individuals diagnosed with ASD, was also classified as “high quality.

Social and Emotional Stimuli

Twenty-six studies assessing PR to social (73.1%) and emotional (26.9%) stimuli, including social interaction, are presented in Table 3. Of the controlled studies comparing reactivity to social stimuli, it was possible to distinguish participants with autism from controls in 12 of 17 studies (70.6%) (Corbett, Schupp, Simon, Ryan, & Mendoza, 2010; Hirstein et al., 2001; Joseph, Ehrman, McNally, & Keehn, 2008; Kylliainen & Hietanen, 2006; Kylliainen et al., 2012; Mathersul, McDonald, & Rushby, 2013; Naber et al., 2007; Riby, Whittle, & Doherty-Sneddon, 2012; Schupp, Simon, & Corbett, 2013; Sheinkopf, Neal-Beevers, Levine, Miller-Loncar, & Lester, 2013; Stagg, Davis, & Heaton, 2013; van Hecke et al., 2009). Across 22 physiological measures employed, participants with autism showed similar PR to controls in respiratory sinus arrhythmia, EDA, and HR (40.9%) when exposed to eye gaze stimuli, facial stimuli with neutral expressions, the strange situation procedure, conversation, and child-directed speech (Mathersul et al., 2013; Naber

et al., 2007; Kaartinen et al., 2012; Klusek, Martin, & Losh, 2013; Louwerse et al., 2013; Watson, Roberts, Baranek, Mandulak, & Dalton, 2012; Willemsen-Swinkels, Bakermans-Kranenburg, Buitelaar, van Ijzendoorn, & van Engeland, 2000); increased PR in cortisol and EDA (9.1%) when exposed to play with peers or eye gaze stimuli (Corbett et al., 2010; Joseph et al., 2008); decreased PR in EDA and cortisol (13.6%) when exposed to human faces, eye gaze stimuli, and the strange situation procedure (Hirstein et al., 2001; Naber et al., 2007; Stagg et al, 2013); different patterns of cortisol reactivity by age (4.5%) when exposed to peer interaction (Schupp et al., 2013); and differential responding to stimuli (22.7%), or non-differential responding (4.5%), to stimuli observed among control participants in EDA, HR and respiratory sinus arrhythmia when exposed to various eye gaze stimuli, faces, a stranger approach procedure, and videos of familiar or unfamiliar persons (Kylliainen & Hietanen, 2006; Kylliainen et al., 2012; Riby et al., 2012; Sheinkopf et al., 2013; van Hecke et al., 2009). One study (4.5%; Mathersul et al., 2013) identified abnormalities in physiological habituation to the stimuli among participants with ASD when exposed to facial stimuli. In one study reviewed (Stagg et al., 2013), participants with autism only had weaker electrodermal reactivity to eye gaze stimuli if they had a co-morbid language delay while participants with autism and no language delay were similar in PR to typically developing controls.

Table 3

Summary of Studies utilising Social or Emotional Stimuli

Study	n	Age range (mean)	Experimental Group Diagnoses	Control Group	Stimuli	Physiological measure(s)	Findings
Ben Shalom et al. (2006)	20	9-18 years (12.4 years)	High-functioning autism; Asperger syndrome	Typically developing	Pictures from the International Affective Picture System	EDA	The groups did not differ significantly in EDR to stimuli.
Blair (1999)	40	- (9.1 years)	Autism	Typically developing	Distress cue stimuli; Threatening stimuli; Neutral stimuli (International Affective Picture System)	EDA	There was no main effect of group on EDR. However, children with autism showed significantly weaker PR to threatening stimuli than control participants.
Bölte et al. (2008)	20	- (27.2 years)	High-functioning autism	Typically developing	Pictures from the International Affective Picture System	HR; Blood pressure	The autism group presented with a stable to increasing pattern of PR to stimuli while the opposite pattern was observed in the control group. Further, the autism group differed from controls in their HR and blood pressure responses to several of the stimuli presented.
Cohen et al. (2015)	40	10-17 years (12.8 years)	Autism; Autism and co-morbid Fragile X syndrome	Typically developing	Emotionally evocative images (NimStim facial Affect Set); Nonsocial pictures	EDA; HR; HRV; Interbeat interval; Vagal tone	The ASD and Fragile X group had significantly higher EDA at baseline than the other groups; EDA and HR responses across stimuli differed very little between groups although within group analyses revealed differential responding according to picture type in electrodermal magnitude in the ASD group and in the number of electrodermal responses of the ASD and Fragile X group according to stimulus type.
Corbett et al. (2010)	45	8-12.5 years (9.9 years)	High-functioning autism	Typically developing	Peer interaction playground paradigm	Salivary cortisol	Among participants with autism only, age mediated cortisol responsivity such that older children had significantly greater reactions to peer interaction than younger children. Significantly more children with autism showed heightened cortisol reactivity to play; Social functioning (Social Communication Questionnaire and Social Responsiveness Scale) was not predictive of cortisol reactivity. PR was positively correlated with peripheral play and negatively correlated with use of gestures.

Hirstein et al. (2001)	50	3-13 years -	Autism	Typically developing	Human faces; Objects	EDA	Controls showed greater EDR to faces while those with autism showed similar reactivity to faces and objects.
Hubert et al. (2009)	32	- (26.4 years)	High-functioning autism; Asperger syndrome	Typically developing	Human faces expressing emotion and neutral faces; Objects	EDA	EDR of participants with autism was significantly weaker than that of controls in response to faces expressing emotion. EDR to neutral faces or objects was similar in both groups; EDR was not correlated with cognitive ability.
Joseph et al. (2008)	40	8.6-15.8 years (12.2 years)	Autism; PDD-NOS	Typically developing	Eye gaze stimuli (direct and averted)	EDA	Children with autism showed stronger EDR to both sets of stimuli; Facial recognition accuracy was negatively correlated with EDR to eye gaze.
Kaartinen et al. (2012)	31	8.5-15.9 years (12.5 years)	Autism; Asperger Syndrome	Typically developing	Eye gaze stimuli (direct, averted and closed eyes)	EDA	There were no inter-group differences in EDR to any of the stimuli; EDR to direct eye gaze was correlated with social communication skills and the use of language, the use of gesture, and nonverbal play (Developmental, Dimensional, and Diagnostic Interview).
Klusek et al. (2014)	68	4.1-14.6 years (9.6 years)	Autism	Typically developing	Conversation	HR; RSA; Interbeat interval; Vagal tone	Participants with ASD showed similar HR and RSA activity to typically developing controls in response to conversation; Neither autism severity nor anxiety (Child Behaviour Checklist) were associated with any of the physiological responses of the ASD group; Neither HR nor interbeat interval change were significantly associated with pragmatic skills (Comprehensive Assessment of Spoken Language). RSA during conversation was a significant predictor of pragmatic skills on one measure (Pragmatic Rating Scale- School Age) but not another (Comprehensive Assessment of Spoken Language).
Kylliainen and Hietanen (2006)	24	6.1-16 years (9.4 years)	Autism	Typically developing	Eye gaze stimuli (direct and averted)	EDA	Participants with autism showed significantly greater EDR to direct eye gaze than averted eye gaze while controls responded the same to both stimuli.
Kylliainen et al. (2012)	29	10.6-14.8 years (12.9 years)	Autism	Typically developing	Eye gaze stimuli (eyes shut, eyes normally open, eyes wide open)	EDA; EEG	EDR to open eyes was significantly greater than to closed eyes among the autism group. Eyes wide open provoked significantly greater EDR than eyes normally open among participants with autism. EDR of control participants was similar for all stimuli.

Lopata et al. (2008)	33	6-13 years (9.8 years)	Asperger syndrome; High-functioning autism; PDD-NOS	-	Interaction with familiar peer; Interaction with unfamiliar peer	Salivary Cortisol	PR to unfamiliar peer interaction was significantly higher than PR to familiar peer interaction. Participants who experienced familiar peer interaction first showed significantly greater PR to the subsequent unfamiliar peer interaction than participants who experienced unfamiliar peer interaction first; Self-reported distress was positively correlated with cortisol reactivity.
Louwerse et al. (2013)	65	12-21 years (16 years)	High-functioning autism	Typically developing	Eye gaze stimuli (direct, averted, or closed eyes)	HR; EDA	The groups did not differ on their HR or EDR to any of the stimuli; Neither HR reactivity nor EDR were correlated with social deficits (Children's Social behaviour Questionnaire).
Louwerse et al. (2014)	73	- (16.1 years)	High-functioning autism	Typically developing	Emotional stimuli with social content or non-social content (International Affective Pictures System)	HR; EDA	The groups did not differ in their electrodermal or HR response to either type of stimulus.
Maras et al. (2012)(i)	38	- (36.5 years)	Autism	Typically developing	Emotional or neutral slide sequence	EDA	It was not possible to differentiate the groups on the basis of PR to either slide sequence.
Maras et al. (2012)(ii)	48	- (41.9 years)	Autism	Typically developing	Emotional or neutral video	HR	HR reactivity to the videos did not differ between the groups.
Mathersul et al. (2013)	61	18-73 years (39.9 years)	High-functioning autism	Typically developing	Facial stimuli with neutral expressions.	EDA; HR; Evoked cardiac deceleration	Cardiac responses to the stimuli were similar in both groups. The groups did not differ in initial EDR to the stimuli. However, abnormalities in habituation to the stimuli were evident in the electrodermal response of autism group.
Naber et al. (2007)	52	- (2.4 years)	Autism; PDD-NOS	Typically developing	Strange situation procedure	HR; Salivary cortisol	Baseline cortisol levels and rhythms did not differ between the groups; Autism did not predict HR reactivity but autism symptoms did predict lower cortisol reactivity.

Riby et al. (2012)	24	12.6-17.6 years (14 years)	Autism	Typically developing	Human face seen on video; Human face seen in person	EDA	While controls showed significantly greater EDR to the faces seen in person, participants with autism showed similar EDR to both types of stimuli.
Schupp et al. (2013)	52	8-12 years (10.1 years)	Autism; PDD-NOS	Typically developing	Peer interaction playground paradigm	Salivary cortisol	Children with ASD had higher average cortisol levels than typically developing children of the same age. For younger children these declined over time while older children with autism were more likely to show cortisol increases in response to the play paradigm.
Sheinkopf et al. (2013)	23	2.5-6.6 years (4.1 years)	Autism	Typically developing	Stranger approach procedure (distal stranger and proximal stranger)	HR; RSA	HR was similar in both groups at baseline; The autism group was significantly more likely to respond physiologically to the proximal stranger than the distal stranger, a relationship which was not observed among controls; PR was associated with social functioning (Vineland Adaptive Behaviour Scales) among participants with ASD.
Stagg et al. (2013)	50	7-15 years (9.8 years)	High-functioning autism-language normal; High-functioning autism-language delay	Typically developing	Eye gaze stimuli (direct, averted, and closed eyes)	EDA	Both typically developing participants and participants with high-functioning autism and normal language presented with significantly stronger PR to the stimuli than participants with high-functioning autism and a language delay. The groups did not differ in their patterns of PR to the different forms of eye gaze stimuli; A positive correlation between verbal mental age (The British Vocabulary Picture Scale-II) and PR to the presented stimuli was identified while neither the onset of first word nor the language and social communication subscale of the Developmental, Dimensional and Diagnostic Interview were correlated with EDR.
van Hecke et al. (2009)	33	8-12 years (9.9 years)	High-functioning autism	Typically developing	Video of familiar person; Video of unfamiliar person	RSA; EEG	Children with autism showed significantly decreased HR regulation in response to the unfamiliar person while controls did not; Higher levels of HR regulation were correlated with better social behaviours and fewer problem behaviours (Social Skills Rating System - Elementary Parent Form; Social Responsiveness Scale-Parent Form).
Watson et al. (2010)	22	2.3-3.5 years (2.9 years)	Autism	-	Child-directed speech; Non-social stimuli	RSA	PR to child-directed speech was positively correlated with receptive language skills at entry and expressive language and social communicative abilities at follow-up (MacArthur-Bates Communicative Developmental Inventory, Words and Gestures Form; The Preschool Language Scale, Fourth Edition; The Mullen Scales of Early Learning; The Vineland Adaptive Behaviour Scales).

Watson et al. (2012)	51	.5-3.5 years (2.4 years)	Autism	Typically developing	Child-directed speech; Non-social stimuli	RSA; Heart period/ Interbeat interval	Participants with autism had smaller interbeat-intervals (indicative of faster HR) in response to both sets of stimuli but did not differ in their RSA response.
Willemsen-Swinkels et al. (2000)	51	- (5.2 years)	Autism; PDD-NOS	Typically developing	Strange situation procedure	HR	Attachment style, rather than diagnosis, predicted HR responses to separation from, and reunion with, parents.

EDA = Electrodermal Activity, EDR = Electrodermal Reactivity, HR = Heart Rate, HRV = Heart Rate Variability, PR = Physiological Reactivity, RSA= Respiratory Sinus Arrhythmia.

Fifteen of the included studies (Corbett et al., 2010; Kylliainen & Hietanen, 2006; Kylliainen et al., 2012; Mathersul et al., 2013; Naber et al., 2007; Riby et al., 2012; Schupp et al., 2013; Sheinkopf et al., 2013; Stagg et al., 2013; van Hecke et al., 2009; Louwerse et al., 2013; Watson et al., 2012; Willemsen-Swinkels et al., 2000; Lopata, Volker, Putnam, Thomeer, & Nida, 2008; Watson, Baranek, Roberts, David, & Perryman, 2010) examining PR to social stimuli were classified as being of higher quality. The exclusion of the remaining studies, did not greatly impact upon the percentage of controlled studies suggesting significant differences in PR between participants with ASD and controls (69.2% from 70.6%).

Of the seven controlled studies comparing reactivity to emotional stimuli, it was possible to distinguish participants with autism from controls (57.1% reporting significant differences) in four studies when exposed to threatening stimuli (e.g., a shark, a gun, a face with an angry expression), pictures from the International Affective Picture System, experimenter affect, emotionally evocative images, and emotional facial expressions (Blair, 1999; Bolte, Feineis-Matthews, & Poustka, 2008; Cohen, Masyn, Mastergeorge, & Hessl, 2015; Hubert et al., 2009). Across 12 physiological measures, participants with autism were similar to controls in HR, HRV, and EDA (58.3%) when exposed to social and nonsocial pictures from the International Affective Picture System, emotional or neutral slides and videos, emotionally evocative images, and nonsocial pictures (Cohen et al., 2015; Ben Shalom et al., 2006; Louwerse et al., 2014; Maras, Gaigg, & Bowler, 2012); demonstrated decreased PR in EDA (8.3%) when exposed to emotional facial expressions (Hubert et al., 2009); a different pattern of PR in HR and BP (16.6%) when exposed to pictures from the International Affective Picture System and different HR and BP responses to certain stimuli (Bolte et al., 2008); differential responding to emotionally evocative images and nonsocial pictures in EDA (8.3%; Cohen et al., 2015); and non-

differential responding in EDA to stimuli (8.3%) than was observed among control participants, when exposed to distressing, threatening or neutral stimuli (Blair, 1999).

Of the three studies (Blair, 1999; Ben Shalom et al., 2006; Louwerse et al., 2014) examining PR to emotional stimuli which were classified as being of higher quality, only one of these (33.3%, a reduction from 57.1%; Blair, 1999) reported differences between individuals with ASD and controls in the form of non-differential responding to stimuli among participants with ASD than was observed among control participants.

Stressor Stimuli

Sixteen studies assessing PR in response to stressor stimuli are presented in Table 4. It was possible to distinguish participants with autism, and participants with autism and co-morbid anxiety, from controls in 10 of 14 studies (71.4%) (Corbett et al., 2006; Goodwin et al., 2006; Hollocks, Howlin, Papadopoulous, Khondoker, & Simonoff, 2014; Jansen, Gispen-de Wied, van der Gaag, & van Engeland, 2003; Jansen et al., 2006; Kushki et al., 2013; Lanni et al., 2012; Levine et al., 2012; Spratt et al., 2012; Tordjman et al., 2009). Across 20 physiological measures, participants with autism responded similarly to controls in cortisol, HR, HRV, and EDA (45%) when exposed to nonsocial environmental stressors (e.g., mock MRI scan), psychosocial stressors (e.g., public speaking task, Trier Social Stress test), and a blood draw stressor (Hollocks et al., 2014; Jansen et al., 2003; Jansen et al., 2006; Levine et al., 2012; Corbett, Mendoza, Wegelin, Carmean, & Levine, 2008; Corbett et al., 2009; Corbett, Schupp, & Lanni, 2012; Rattaz et al., 2013); demonstrated increased PR in cortisol and HR (25%) when exposed to a nonsocial environmental stressor, a psychosocial stressor, an anxiety inducing task. and blood draw stressors (Corbett et al., 2006; Hollocks et al., 2014; Kushki et al., 2013; Spratt et al., 2012; Tordjman et al., 2009); decreased PR in HR, cortisol and EDA (25%) when exposed to common stressors (derived from the Stress Survey Schedule for Individuals with Autism and other Developmental Disabilities), psychosocial stressors, and an anxiety-inducing

task (Goodwin et al., 2006; Jansen et al., 2006; Kushki et al., 2013; Lanni et al., 2012; Levine et al., 2012); and different patterns of PR in HR (5%) when exposed to psychosocial stressors (Jansen et al., 2003). Four studies (Spratt et al., 2012; Tordjman et al., 2009; Corbett et al., 2012; Rattaz et al., 2013) noted a prolonged duration for physiological recovery, or higher physiological activity during the recovery period, among participants with autism, as compared to controls, following exposure to a nonsocial environmental stressor, psychosocial stressors or blood draw stressors. Hollocks et al. (2014) exposed participants with autism, autism and a co-morbid anxiety disorder, and typically developing controls to an adapted version of the Trier Social Stress Test. They found that participants with autism alone had the highest HR response to the stressor while participants with autism and a co-morbid anxiety disorder were distinguishable from controls and participants with ASD alone on the basis of a blunted cortisol response to the stressor and slower HR recovery.

Table 4

Summary of Studies utilising Stressor Stimuli

Study	n	Age range (mean)	Experimental Group Diagnoses	Control Group	Stimuli	Physiological measure(s)	Findings
Corbett et al. (2006)	22	6-11 years (8.8 years)	Autism	Typically developing	Nonsocial environmental stressor (mock MRI scan)	Salivary Cortisol	Cortisol circadian rhythms were similar among both groups. However, there was greater variability in cortisol levels across the day in the autism group; The majority of participants with ASD showed cortisol increases in response to the stressor while the majority of controls showed no response or a decrease in cortisol; There was no relationship between scores on the Stress Survey Schedule or Short Sensory Profile and cortisol.
Corbett et al. (2008)	44	6.5-12 years (9.1 years)	Autism	Typically developing	Nonsocial environmental stressor (mock MRI scan and real MRI scan)	Salivary cortisol	Both groups showed the expected cortisol rhythms although, over the course of sampling, morning cortisol levels decreased in the autism group and evening cortisol levels were consistently higher than in controls. Greater variability in cortisol levels was also observed in the autism group; The groups did not differ in PR to the initial stressor and both demonstrated increased PR prior to the second stressor presentation.
Corbett et al. (2009)	44	6-12 years (9.1 years)	Autism	Typically developing	Nonsocial environmental stressor (mock MRI scan)	Salivary Cortisol	There was greater variability, and dysregularity, in cortisol levels throughout the day in the autism group than in the control group; The groups did not differ in cortisol reactivity to the stressor; PR to the stressor was not significantly related to Stress Survey Schedule or Short Sensory Profile Score.

Corbett et al. (2012)	59	8-12.6 years (10 years)	Autism; PDD-NOS	Typically developing	Trier Social Stress Test- Child version; The Peer Interaction Playground Paradigm	Salivary Cortisol	PR to the stressors did not differ between the groups. However, significantly greater variability in cortisol levels across the stressor paradigms was noted in the autism group; Quick physiological recovery from stress was evident in the typically developing group while the physiological response in the autism group persisted for significantly longer.
Goodwin et al. (2006)	10	8-18 years (13.8 years)	Autism	Typically developing	Stressors from the Stress Survey Schedule for Persons with Autism and other Developmental Disabilities	HR	Baseline HR was higher among participants with autism than among typically developing controls; The autism group demonstrated significant PR to stressors on 22% of occasions while the typically developing participants showed significant PR on 60% of occasions.
Groden et al. (2005)	10	13-27 (24 years)	Autism; PDD-NOS	-	Stressors from the Stress Survey Schedule for Persons with Autism and other Developmental Disabilities	HR	Each of the stressor domains led to significant HR changes in some participants. However PR, and the stressors than evoked it, demonstrated a high degree of individual variability.

Hollocks et al. (2014)	75	10-16 years (13.2 years)	Autism; Autism and co-morbid anxiety disorder	Typically developing	Trier Social Stress Test	HR; HRV; Salivary Cortisol	The ASD group had a higher resting HR than the ASD and anxiety group and controls while the groups did not differ on baseline measures of HRV or cortisol; The groups did not differ on HRV response to the stressor. The ASD group had higher HR during the stressor than the other groups while the ASD and anxiety group showed lower cortisol reactivity to the stressor than the other groups; Participants with ASD and controls did not differ significantly in cortisol recovery slope while the HR of those with ASD and anxiety was slower to recover; HR reactivity and cortisol reactivity were negatively correlated with anxiety for the ASD and anxiety group only (The Child and Adolescent Psychiatric Assessment; The Spence Children's Anxiety Scale).
Jansen et al. (2003)	22	- (9.4 years)	Autism	Typically developing	Psychosocial stressor	HR; Salivary Cortisol	Children with autism did not differ from controls in their cortisol response to the psychosocial stressor although they showed a different pattern of HR responses; There was no correlation PR and scores on the Child Behaviour Checklist or IQ. Communication scores on the Autism Diagnostic Interview were correlated with cortisol during the stressor.
Jansen et al. (2006)	24	- (21.3 years)	Autistic disorder; Asperger syndrome	Typically developing	Psychosocial Stressor	HR; Salivary Cortisol; Adrenocorticotropic hormone; Oxytocin; Vasopressin; Norepinephrine; Epinephrine	The autism group showed a decreased HR response to the stressor while their neuroendocrine response did not differ from that of controls; HR reactivity to the stressor was positively correlated with social interaction and communication (Autism Diagnostic Interview); The decreased HR reactivity was associated with less self-care and greater current social anxiety (Dutch Social Interaction Inventory).

Kushki et al. (2013)	29	8-15 years (11.1 years)	Autism; Asperger syndrome	Typically developing	Anxiety-inducing task	HR; EDA; Skin temperature	The autism group had significantly elevated HR and EDA during baseline; Participants with autism demonstrated higher HR in response to the stimulus than controls while participants with autism demonstrated a blunted electrodermal response to the stressor.
Lanni et al. (2012)	30	8-12 years (9.7 years)	Autism	Typically developing	Trier Social Stress Test- Child Version	Salivary cortisol	There were no significant differences in resting cortisol levels between the groups; While control participants showed a greater cortisol response to the stressor, between group differences were not significant; Verbal ability (Verbal Fluency Test) was not related to cortisol responses. Self-reported anxiety (State-Trait Anxiety for Children) was also unrelated to cortisol responses.
Levine et al. (2012)	30	8-12 years (9.7 years)	High-functioning autism; Asperger syndrome; PDD-NOS	Typically developing	Trier social stress test	Salivary Cortisol; EDA; Vagal tone	Neither cortisol, EDA, nor vagal tone differed between the groups at baseline; Individuals with autism were significantly more likely to show cortisol decreases in response to the stressor while controls typically showed cortisol increases. There were no differences in EDR in response to the stressor between the groups; The groups did not differ significantly on any of the physiological measures during the recovery period.
Moskowitz et al. (2013)	3	6-9 years (7.7 years)	Autism spectrum disorder	-	Exposure to high anxiety context	HR; RSA	For two participants, HR was significantly higher during the high anxiety context than during a low anxiety context while RSA was also lower during the high-anxiety context for two participants; All participants presented with more anxious behaviour and more challenging behaviour during the high anxiety condition.

Rattaz et al. (2013)	71	- -	Autism spectrum disorder	Typically developing	Blood draw stressor	HR	Children with autism had higher HR at baseline than controls; Participants with autism did not differ from controls in their HR reactivity to the stressor; The heart rate of children with autism remained higher during the recovery period than that of controls.
Spratt et al. (2012)	48	3-12 years (6.1 years)	Autism	Typically developing	Blood draw stressor; Novel environment	Urinary, serum, and salivary cortisol	Children with ASD had significantly higher serum cortisol levels at baseline; Children with autism showed a significantly greater cortisol response to the stressor; Children with autism showed a prolonged cortisol response duration; There were no correlations between autism severity or symptoms (CARS score, DSM IV scores) or behaviour (The Child Behaviour Checklist) and any of the cortisol measures.
Tordjman et al. (2009)	188	- (12.3 years)	Autism	Typically developing	Blood draw stressor	HR; Plasma β-Endorphin	The autism group had a significantly higher HR during baseline; The autism group showed greater HR reactivity to the stressor; The autism group had a significantly higher HR after the stressor.

EDA = Electrodermal Activity, EDR = Electrodermal Reactivity, HR = Heart Rate, HRV = Heart Rate Variability, PR = Physiological Reactivity, RSA= RSA.

The examination of the 12 studies categorised as being of "high" quality (Corbett et al., 2006; Corbett et al., 2009; Corbett et al., 2012; Goodwin et al., 2006; Groden et al., 2005; Hollocks et al., 2014; Jansen et al., 2003; Jansen et al., 2006; Lanni et al., 2012; Levine et al., 2012; Mozkowitz et al., 2013; Tordjman et al., 2009) revealed that the exclusion of studies of lower quality led to a somewhat increased percentage of studies (80% up from 71.4%) reporting discernible differences in physiological response between participants with autism and controls. This process also eliminated two of the four studies (Spratt et al., 2012; Rattaz et al., 2013) reporting a prolonged stress response among individuals with ASD.

Relation between Behavioural or Psychological Variables and Physiological Reactivity

The reviewed studies were also examined for evidence of consideration of behavioural or psychological variables that may be associated with PR. In total, 26 of the studies reviewed (45.6%; (Chang et al., 2012; Corbett et al., 2006; Corbett et al., 2009; Corbett et al., 2010; Hollocks et al., 2014; Hubert et al., 2009; Jansen et al., 2003; Jansen et al., 2006; Joseph et al., 2008; Kaartinen et al., 2012; Klusek et al., 2014; Lanni et al., 2012; Lopata et al., 2008; Louwerse et al., 2013; Moskowitz et al., 2013; Naber et al., 2007; McCormick et al., 2014 Schaaf et al., 2015; Schoen et al., 2009; Sheinkopf et al., 2013; Spratt et al., 2012; Stagg et al., 2013; van Engeland et al., 1991; van Hecke et al., 2009; Watson et al., 2010; Woodard et al., 2012) examined the association between PR and psychological or behavioural variables. The findings of individual studies with regards the correlation between PR and such variables are outlined in Tables 2, 3, and 4. It was most common for studies to assess the association between PR and measures of social and communication abilities such as the Autism Diagnostic Inventory, Developmental, Dimensional, and Diagnostic Interview, Children's Social Behaviour Questionnaire, Vineland Adaptive Behaviour Scale, Social Communication Questionnaire, Social

Responsiveness Scale, Social Skills Rating System, MacArthur-Bates Communicative Developmental Inventory, Words and Gestures Form, and behavioural observations of play or use of gestures, (34.6%); (Corbett et al., 2010; Jansen et al., 2003; Jansen et al., 2006; Kaartinen et al., 2012; Louwerse et al., 2013; Sheinkopf et al., 2013; Stagg et al., 2013; van Hecke et al., 2009; Watson et al., 2010). With the exception of Louwerse et al. (2013) and Stagg et al. (2013), all other studies reported significant correlations between PR to stimuli and at least one measure relating to social or communicative abilities. The relation between PR and sensory behaviours was examined in six studies (23.1%; (Chang et al., 2012; Corbett et al., 2006; Corbett et al., 2009; Legisa et al., 2013; Schoen et al., 2009; Woodard et al., 2012). Findings regarding the correlation of PR with measures of sensory behaviours including the Short Sensory Profile, the Sensory Processing Measure, and the Infant /Toddler Sensory Profile, were mixed with two studies (33.3%) reporting some correlation and four studies reporting no discernable correlation between these variables. The correlation between PR and indirect measures of stress or anxiety, such as the Stress Survey Schedule for Persons with Autism and other Developmental Disabilities, The Child and Adolescent Psychiatric Assessment, The Child Behaviour Checklist, The Spence Children's Anxiety Scale, The Dutch Social Interaction Inventory, State-Trait Anxiety for Children, and self-reported distress, was examined by seven studies (26.9%; Corbett et al., 2006; Corbett et al., 2009; Hollocks et al., 2014; Jansen et al., 2006; Klusek et al., 2014; Lanni et al., 2012; Lopata et al., 2008) with only two (28.6%) of these reporting the identification of a correlation between these variables. Seven studies (26.9%; (Hubert et al., 2009; Jansen et al., 2003; Klusek et al., 2014; Lanni et al., 2012; Stagg et al., 2013; van Engeland et al., 1991; Watson et al., 2010) examined the correlation between PR and measures of intellectual or verbal functioning, such as the Comprehensive Assessment of Spoken Language, the Expressive Vocabulary Test, Pragmatic Rating Scale, Verbal Fluency Test, the Mullen Scales of Early Learning, MacArthur-Bates Communicative

Developmental Inventory, Words and Gestures Form, The Preschool Language Scale, verbal mental age as per the British Vocabulary Picture Scale, and IQ, with only three of these studies (42.9%; (Stagg et al., 2013; Klusek et al., 2014; Watson et al., 2010) suggesting any correlation between PR and intellectual functioning or verbal ability. Four studies (16%; Jansen et al., 2013; McCormick et al., 2014; Spratt et al., 2012; van Hecke et al., 2009) examined the correlation between PR and indirect measures of behaviour, including the Child Behaviour Checklist, the Repetitive-Behaviour Scale-Revised, Social Skills Rating System, with only one of these studies (25%; van Hecke et al., 2009) suggesting a relation between PR and behaviour. Two studies (7.7%) examined the relation between PR and observations of challenging behaviour (Moskowitz et al., 2013) and behavioural reactivity to stimuli (Schaaf et al., 2015), with both suggesting a relation between overt behaviour and physiological responses. Finally, three studies (11.5%; Naber et al., 2007; Klusek et al., 2014; Spratt et al., 2012) assessed the association between autism symptoms or severity, as measured by the Childhood Autism Rating Scale (CARS), Diagnostic and Statistical Manual of Mental Disorders IV (DSM) Scores, and ADOS, with only one of these (Naber et al., 2007) suggesting any relation between PR and autism severity. Additionally, three studies (Cohen et al., 2015; Hollocks et al., 2014; Stagg et al., 2013) examined the impact of a co-morbid diagnosis or additional impairment, fragile x syndrome, a language delay, and an anxiety disorder respectively, on PR among those with ASD. In each of these studies physiological activity differed between those with a sole diagnosis of autism and those with a co-morbid diagnosis or additional delay. Finally, Jansen et al (2006) reported a correlation between PR to a stressor and self-care, and Joseph et al. (2008) reported a correlation between PR and facial recognition accuracy.

The exclusion of studies found to be of lower quality led to the removal of five studies (Chang et al., 2012; Hubert et al., 2009; Joseph et al., 2008; Klusek et al., 2014; Spratt et al., 2012). This resulted in a reduction in the body of evidence suggestive of a

relationship between PR and sensory behaviours, of an association between PR and facial recognition accuracy, PR and verbal or intellectual ability. The one remaining study examining autism symptoms suggested a positive association with PR. The exclusion of these studies did not reduce the evidence suggestive of the association between PR and indirect or direct measures of behaviour, indirect measures of anxiety or stress, social and communication ability, or the impact of various co-morbid diagnoses on PR among those with ASD.

Baseline Physiological Arousal

Previously mentioned theories of chronic hyper- and hypo-arousal among individuals with autism also prompted us to examine baseline levels of physiological arousal reported by the studies included in the review. Of the 22 studies reporting baseline physiological activity across 30 physiological measures, participants with autism and controls did not differ on 50% of these baseline measures, including cortisol, HR, HRV, respiratory sinus arrhythmia, and EDA (Cohen et al., 2015; Corbett et al., 2006; Hollocks et al., 2014; Lanni et al., 2012; Levine et al., 2012; McCormick et al., 2014; Naber et al., 2007; Palkovitz & Wiesenfeld, 1980; Sheinkopf et al., 2012; van Engeland, 1984; Zahn et al., 1987); participants with ASD were more physiologically aroused on 40% of measures, including cortisol, EDA, HR, HRV and respiratory sinus arrhythmia (Chang et al., 2012; Goodwin et al., 2006; Hollocks et al., 2014; Kushki et al., 2013; Palkovitz & Wiesenfeld, 1980; Rattaz et al., 2013; Schaaf et al., 2015; Schupp et al., 2013; Spratt et al., 2012; Tordjman et al., 2009; Zahn et al., 1987); lower on one (3.3%) measure of EDA (Schoen et al., 2009); and showing differences in cortisol secretion across the day in two studies (6.7% of measures; (Corbett et al., 2008; Corbett et al., 2009). One non-controlled study employing a measure of EDA when exposed to the sensory challenge protocol (Schoen et al., 2008) identified both patterns of basal hypo-arousal and hyper-arousal among participants with autism. Furthermore, one study (Cohen et al., 2015) noted differences in

the baseline physiological arousal of those with ASD and co-morbid Fragile X syndrome as compared to participants with ASD alone and typically developing controls.

The exclusion of lower quality studies upon findings regarding baseline physiological activity appeared to have minimal impact on the overall pattern of results obtained. Of the 14 remaining studies of higher quality which employed 18 physiological measures (Corbett et al., 2008; Corbett et al., 2009; Goodwin et al., 2006; Hollocks et al., 2014; Lanni et al., 2012; Levine et al., 2012; McCormick et al., 2014; Naber et al., 2007; Schaaf et al., 2015; Schoen et al., 2009; Schupp et al., 2013; Sheinkopf et al., 2013; Tordjman et al., 2009; Zahn et al., 1987), there were no significant differences in baseline physiological arousal on 10 measures (55.6%; (Corbett et al., 2006; Hollocks et al., 2014; Lanni et al., 2012; Levine et al., 2012; McCormick et al., 2014; Naber et al., 2007; Sheinkopf et al., 2013; Zahn et al., 1987), higher arousal among participants with ASD on five measures (27.8%; Goodwin et al., 2006; Hollocks et al., 2014; Schaaf et al., 2015; Schupp et al., 2013; Tordjman et al., 2009), lower arousal among participants with ASD on two measures (11.1%; Schoen et al., 2009; Zahn et al., 1987), and one study suggested differences in the daily patterns of cortisol secretion of those with ASD (5.6%; Corbett et al., 2009).

Discussion

The current systematic review examined PR to sensory, social and emotional, and stressor stimuli among individuals with autism across 57 studies published between the years of 1978 and 2014. Findings suggest abnormalities in PR exist for at least some individuals with autism. Differences in PR on at least one physiological measure were observed in 78.6% of studies employing sensory stimuli; 66.7% of studies employing social or emotional stimuli; and 71.4% of studies employing stressor stimuli. Notably, however, findings were not uniform, even when the same or very similar physiological measures, test stimuli, methodological protocols, and sample characteristics were used.

Inconsistencies in results across these studies are an important finding of this review, and raise questions about whether atypical PR observed in individuals with autism is a valid marker for the existence of clinical subtypes, relate to clinically relevant behaviours, or arise from methodological artifacts.

Given the highly heterogeneous presentation of autism (Stagg et al., 2013), it is perhaps unsurprising that PR varies widely across individuals who share the same diagnosis. The outcomes of the quality assessment included here suggest that the differences in PR, and inconsistent outcomes, persist even among the studies that were classified as being of higher quality. The purpose of the assessment of the methodological rigor of the included studies was to determine whether the methodological quality of the studies included impacted upon the outcomes of these studies. To achieve this, we assessed each study on a number of indicators of methodological strength and computed scores that allowed us to assign studies to a “low” or “high” quality category. The exclusion of the “low” quality studies resulted in limited impact on the overall outcomes of each category, or the great degree of inconsistency observed in each stimulus category. Therefore, the results allow us to conclude, with a greater degree of certainty, that PR to the various stimuli examined was variable among persons with autism.

A potential explanation for the high degree of variability in PR observed is the possible existence of subtypes of ASD characterised by different physiological profiles. The putative existence of subtypes within the autism spectrum is not new (Piggott, 1979), and may account for the high degree of variability in PR observed among participants with autism in a number of the studies reviewed (Allen et al., 2013; Corbett et al., 2006; Corbett et al., 2008; Corbett et al., 2012; Groden et al., 2005). Arguably the best current evidence for this hypothesis comes from the studies by Hirstein et al. (2001) and Schoen et al. (2008) who found distinct subtypes of physiological responders in their sample (i.e., hypo-aroused, normally aroused, hyper-aroused) while exposed to the same within-study stimuli

sets. Future research is needed to further examine the prevalence of physiological subtypes among individuals with autism to determine whether such patterns of responding are stable over time, constitute meaningful differences, and have predictive validity.

A number of the studies reviewed also suggest that an association may exist between PR and a number of behaviours and psychological variables highly relevant to individuals with autism, including social and communication abilities, intellectual or verbal functioning, autism symptoms or severity, overt behaviours including both challenging and sensory behaviours, co-morbid diagnoses or delays, and indirect measures of stress or anxiety. Further research is needed to determine the nature of this relationship given its potential clinical significance. For instance, van Hecke and colleagues (2009) found that participants with autism responded to social interactions with substantial increases in physiological arousal, suggesting that the experience was highly stressful for them. The authors suggest that physiological activation in the form of a nervous system primed to fight or flee during such encounters would directly hamper and impede the ability to engage in appropriate social interactions. In other cases, however, the association may not be causal and may be mediated by other variables. For example, Watson et al.'s (2010) finding that increased PR to child-directed speech was positively associated with current and future language and communication skills might be explained by attentional processes. Furthermore, several of the studies reviewed (Chang et al., 2012; Goodwin et al., 2006; Tordjman et al., 2009) suggest that the observable behaviour of an individual with autism may not be indicative of his or her internal physiological arousal state. Baron and colleagues (2006) emphasised the importance of further investigating this phenomenon systematically, especially in individuals with autism who have communication deficits and/or limited emotional expressivity. Future research examining the association between PR and overt behaviour, and the association between PR and important behavioural outcomes, among individuals with autism is warranted. For instance, research on the

Yerkes-Dodson law (1908) has led scholars (Hebb, 1955; Stennett, 1957) to suggest a curvilinear relationship between physiological arousal and performance, wherein performance decreases when arousal is appreciably lower (i.e., an inert effect) or higher (i.e., an anxious effect) than an individual's homeostatic set point. In this way, a certain level of PR to stimuli, or the elevation of physiological arousal in response to stimuli, is necessary for appropriate interactions with a given stimulus. Correspondingly, weak or excessively strong PR may compromise performance or lead to inappropriate reactions to stimuli.

The association between PR and a variety of important health outcomes also suggests value in assessing ANS and LHPA axis functioning in individuals with autism. Negative health outcomes have been linked to both increased and reduced PR (Lovallo, 2011). Much research has evidenced a connection between increased PR to stressors and an increased risk of hypertension and coronary heart disease (Lovallo & Wilson, 1992; Munck, Guyre, & Holbrook, 1984). Although the relationship has received less research attention, Lovallo (2011) described how reduced PR to stressors is likely related to increased risk for autoimmune or metabolic disorders, obesity, and engagement in behaviours with negative health consequences. Individuals with autism were observed to produce statistically significantly greater and weaker PR than controls to a wide variety of stimuli in this review. Assuming PR in an experimental context reflects PR in the natural environment, it is possible that these individuals are at risk for a variety of negative health outcomes due to their physiological over- or under-reactions. Further, the findings of elevated baseline levels of physiological arousal in many of the studies (Chang et al., 2012; Goodwin et al., 2006; Hollocks et al., 2014; Kushki et al., 2013; Palkovitz & Wiesenfeld, 1980; Rattaz et al., 2013; Schaaf et al., 2015; Schupp et al., 2013; Spratt et al., 2012; Tordjman et al., 2009) also suggest that individuals with autism may be at increased risk for negative health outcomes associated with chronic stress, including reduced immune

system responsivity, cardiovascular problems, stunted growth, and diabetes (Romanczyk & Gillis, 2006). Given the serious nature of these health outcomes, longitudinal investigations are warranted to determine if abnormal PR evidenced among individuals with autism can predispose them to ill-health over time.

Finally, studies within this review suggest areas of potential physiological dysregularity in autism that merit future investigations. For instance, the LHPA axis and vagal system were both identified by recent research studies (Corbett et al., 2009; Jansen et al., 2006; Lanni et al., 2012) as being possibly impaired in this population. The LHPA axis plays an important role in stress responses and was previously implicated as a potential underlying cause of autism for some individuals with the disorder (Chamberlain & Herman, 1990). Findings of greater cortisol variability, and atypical cortisol circadian rhythms or levels among individuals with autism (Corbett et al., 2006; Corbett et al., 2008; Corbett et al., 2009; Corbett et al., 2012) in the studies reviewed support the possibility that LHPA axis functioning may be impaired in this population. Jansen et al. (2006) implicated the vagal system as potentially dysfunctional, as participants with autism presented with abnormal HR responses to psychosocial stressors. The vagal system is thought to play a key role in social engagement and communication, and has been posited to relate to diagnostic features associated with autism (Jansen et al., 2006; Porges, 2001). Such suggestions of physiological dysregularity highlight avenues for further research that may contribute to our understanding of the development or the presentation of autism.

The current systematic review was limited in a number of ways. The decision to include all studies that examined PR among individuals with autism, regardless of experimental design, sample size, or year of publication, may be criticised. However, given the breadth and span of research studies published on the topic and lack of a useful quantitative synthesis or overview of these to date, the current review sought to be inclusive and to provide a comprehensive summary and discussion of the current state of

research in this area. Further, many of the included studies that utilised small sample sizes were considered to provide important findings, such as the presence of both hypo- and hyper-arousal among participants with autism (Schoen et al., 2008). Such a study may, for example, contribute to our understanding of discrepant findings observed in many of the other controlled studies and also have important implications for future research including the need to analyse variability in responding among participants with autism and greater consideration of participants' baseline physiological activity. Our division of stimuli into the categories of sensory, social and emotional, and stressor may also be critiqued given the overlap between these classes and our reliance on authors' description of stimuli and their intent of employment for categorisation. However, it was considered necessary to subdivide the articles to better represent the findings with regards PR to the various stimuli and division by stimulus employed was deemed the most meaningful manner of doing this by our research team. Finally, our use of a self-developed, subjective, measure of quality to assess the included studies may also be criticised. However, extensive literature searches did not produce an already existing quality assessment measure suitable for use with the included studies. Given the inconsistent findings of the studies reviewed, it was considered important to assess within the current review whether methodological artifacts may have accounted for the variability in outcomes. Future research could further examine these limitations by utilising more stringent inclusion criteria than those reported here and assessing methodological quality of studies in an alternative manner.

In spite of these limitations, we believe the current review provides an important foundation for future research in this area. First, it is hoped that the provision of explicit indicators of methodological quality will have a positive impact upon future research studies in this area. Outcomes on the quality measure developed for this review were generally quite poor with scores ranging from 1-7 (M=3.8) out of a possible 12 points. Studies were most frequently faulted for the poor consideration of extraneous factors (e.g.,

co-morbid psychological or medical diagnoses, medication usage, affect, physical fitness, physical activity, exposure to novel settings, exposure to novel physiological recording devices) that may impact upon physiological outcomes (i.e., internal validity). Further, less than half of the studies employed more than one physiological measure. The use of multiple indices of PR obtained simultaneously is typically advised, as certain measures can be more sensitive to stimulus perception than others. This phenomenon, referred to as directional fractionation, indicates that autonomic responses may differ in their direction of change in response to external stimulation (Feurstein, Labbé, & Kucmierczyk, 1986). For example, in response to external arousal or unanticipated stimulation, increases in EDA have been frequently accompanied by HR decreases, although covariance of both responses is expected (Hugdahl, 1998). The possibility of such "fractionated" responding underlines the importance of measuring the activation of multiple physiological systems in response to stimulation. For instance, Lovallo (2005) describes a series of experiments in which typically developing individuals engage in tasks that produce either aversive consequences or rewards. Cardiovascular reactivity to both tasks was similar; however, cortisol reactivity differed such that aversive tasks led to cortisol increases and reward tasks did not. Relying on only cardiovascular or cortisol measures in these studies may have failed to appreciate the complex relationship observed. The included studies performed better in the descriptive validity, external validity, and statistical conclusion validity categories of the quality assessment. However, in studies that analysed group responding, the consideration of individual responding was infrequent. This is problematic given the findings of Hirstein et al. (2001) and Schoen et al. (2008) concerning potential subtypes of physiological responders among those with ASD, and the possible impact of individuals presenting with hyper- or hypo-arousal on the outcomes of studies employing solely group analyses. Future research should seek to improve upon the methodological

quality of past studies as a potential means of elucidating the nature of any differences in PR in autism.

The included studies can also be critiqued on a number of additional factors including the absence of screening for maltreatment, a suggested determinant of PR (Carrey, Butter, Persinger, & Bialik, 1995), in all but one study (Spratt et al., 2012), the prevailing focus on higher-functioning individuals with ASD or those with no co-occurring cognitive impairment (although research suggests up to 70% of those with ASD have a co-morbid intellectual disability; La Malfa, Lassi, Bertelli, Salvini, & Placidi, 2004), and the infrequent examination of the impact of co-morbid psychological diagnoses, prevalent among those with ASD (Simonoff et al., 2008), on PR. The role of such factors in PR and autism could be further analysed in future research. This review also highlights the need for a quantitative analysis of findings in this literature and the examination of potential moderator variables, such as age, level of functioning, co-morbid diagnoses, type of stimulus presented, medication status, or physiological measures used. It also suggests numerous avenues for future research including: studies exploring potential subtypes in autism distinguishable by physiological arousal or reactivity and associated profiles or behavioural presentations within these subtypes; examination of cortisol variability or dysregularity among individuals with autism and its impact on overt behaviours; correspondence between PR and overt behaviour or self-reported arousal or stress in autism; and examination of the relationship between PR and health issues in autism. The current findings also highlight the need for greater consideration of participant and stimulus characteristics in future studies of this kind.

Allen et al. (2013, p. 440) conclude that “within-group variability is much larger than any effect of autism, if it exists” and perhaps this can be considered a succinct summary of the findings of the present review. While the majority of studies indicate some abnormalities in PR to sensory, social and emotional, and stressor stimuli among

individuals with autism, other studies have evidenced normal PR to these stimuli. Such discrepant findings indicate that some atypicality exists in PR for some individuals with autism, but not for all. The present review thus underscores the need for future research to investigate such discrepancies. It is likely that further investigation of PR among individuals with autism will further our understanding of the disorder, potential subtypes within it, associated behaviours, and health outcomes.

The outcomes of the systematic review reported in Chapter 2 have provided an important foundation for the empirical research studies that follow. The inconsistencies in the findings of previous psychophysiological research studies in ASD regarding differences in physiological activity between individuals with autism and typically developing persons, the conflicting findings on the relation between physiological activity and behaviour in this population, and the predominant focus of previous research on high-functioning persons with autism, were all considered important factors to consider when conducting research in this area. The current chapter thus prompted us to further explore: levels of physiological activation among a diverse sample of individuals with autism (Experiment 1 of Study 2: Chapter 3); the relationship between physiological activation and engagement in a variety of challenging behaviours among a diverse sample of individuals with ASD (Experiment 2 of Study 2: Chapter 3); and the value of exploring the association between physiological activity and engagement in repetitive, stereotyped behaviour in the context of behavioural assessments of challenging behaviour (Study 3: Chapter 4 and Study 4: Chapter 5).

Chapter 3: Study 2

Salivary Cortisol Levels and Challenging Behaviour in Children with Autism Spectrum Disorder[2]

[2] This chapter has been accepted for publication: Lydon, S., Healy, O., Roche, M., Henry, R., Mulhern, T., & Hughes, B. M. (2015). Salivary cortisol levels and challenging behavior in children with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *10*, 78-92. doi: 10.1016/j.rasd.2014.10.020

Abstract

A relationship between stress and challenging behaviour in individuals with Autism Spectrum Disorder (ASD) has been theorised but infrequently examined empirically. The current study reports two related experiments assessing physiological stress, and its association with challenging behaviour, among persons with ASD. The first experiment compared physiological stress (diurnal salivary cortisol) between a sample of participants with ASD and typically developing peers matched on age, gender, and pubertal status. A small, but non-significant, effect of diagnosis was noted whereby individuals with autism may experience higher stress than their typically developing peers. The second experiment sought to examine the relationship between a parent-reported measure of stress, a physiological measure of stress (diurnal salivary cortisol), and various topographies of challenging behaviour among 61 children and adolescents diagnosed with ASD between the ages of three and 18 years. Differences in cortisol levels (large effect) between those engaging in high and low rates of stereotyped behaviour were observed such that higher levels of stereotypy appeared an overt manifestation of higher levels of stress. The results of the current study suggest that individuals with autism may experience elevated levels of stress and that, for a subset of individuals with ASD, stereotyped behaviour may be an indicator of elevated cortisol levels.

The results of Study 1 (Lydon et al., in press/Chapter 2) indicated that inconsistencies in the findings of psychophysiological research among persons with ASD are prevalent, that the relation between physiological activity and behaviour remains unclear, and that individuals with autism who are low-functioning (IQ<70) have been underrepresented in the psychophysiological research literature. The current research study thus aimed to examine levels of physiological activation among children and adolescents with autism as compared to their typically developing peers and the relationship between physiological activity and engagement in challenging behaviours among a large, diverse sample of children and adolescents diagnosed with ASD. Characterisics of the sample potentially related to physiological activity (e.g., age, level of functioning, medication status) were also explored in this study.

As described in Chapter 1, a great number of theories have posited a link between physiological activity and engagement in challenging behaviour by persons with autism. Empirical studies assessing the association between these variables have primarily used measures of autonomic reactivity such as measures of HR (e.g., Barrera et al., 2007; Lydon et al., 2013; Freeman et al., 1999) and EDA (e.g., Hirstein et al., 2001; Romanczyk & Matthews, 1998). The results of these studies have been mixed with some suggesting that challenging behaviours, which have included stereotypy, SIB, and aggression, are negatively reinforced by the decreases in physiological arousal which they induce (e.g., Barrera et al., 2007) while other suggest that such behaviours are positively reinforced by the increases in physiological arousal observed following their occurrence (e.g., Lydon et al., 2013; Freeman et al., 1999).

Of primary relevance to the current study, are a number of theoretical and experimental papers that have proposed a relationship between stress or anxiety and challenging behaviour among those with developmental disabilities. Groden et al. (1994) theorised that individuals with ASD have a unique vulnerability to stress and anxiety, due

to impairments in social, communicative, cognitive, and sensory functioning, and that these feelings and experiences likely contribute to the symptoms of ASD and engagement in challenging behaviour. The authors further suggest that challenging behaviours may be understood as overt manifestations of internal stress or as a maladaptive coping response. Janssen and colleagues (2002) have proposed a model of challenging behaviour that implicates stress and attachment style in the development of challenging behaviour among individuals with intellectual disabilities. This model, derived from previous research, highlights studies showing that individuals with severe or profound intellectual disabilities are at greater risk of both experiencing stress and of developing poor attachment styles. Previous research has suggested that these two factors in combination may predispose an individual to the development of behavioural problems and this may be the case for some individuals with intellectual disabilties and co-morbid challenging behaviour (Janssen et al., 2002).

A number of studies have empirically examined the relationship between challenging behaviour and physiological measures of stress. The activation of the LHPA axis in response to stress or challenge has been found to result in enhanced release of cortisol, a steroid hormone, from the adrenal glands. Cortisol levels can be assessed readily in several biological fluids including saliva, urine or blood. Elevated levels of cortisol have been found to directly correlate with levels of stress. Verhoeven and colleauges (1999) examined the relationship between baseline levels of various hormones and neurochemicals, including cortisol, and SIB or stereotyped behaviour exhibited by individuals with intellectual disabilities. The authors noted a tendency towards lower values of total cortisol among those engaging in a high level of SIB or a high level of stereotyped behaviour. Symons et al. (2003) compared cortisol levels among individuals with developmental disabilities and chronic, severe SIB and those with developmental disabilities and no SIB. A trend for those with SIB to have higher cortisol levels was

observed. Further, morning and evening cortisol levels were positively correlated with SIB severity. Symons et al. (2011) examined cortisol and salivary α-amylase among 51 adults with intellectual and developmental disabilities, 34 of whom engaged in severe SIB and 17 of whom acted as controls. Cortisol levels were found to be significantly higher among those with SIB than among the matched controls. However, a more recent study by Gabriels and colleagues (2013) examined the relationship between repetitive behaviours and cortisol levels among 21 children with ASD. It was found that participants with high levels of repetitive behaviour had lower levels of cortisol than participants who engaged in low levels of repetitive behaviours. Thus, research suggests that there may exist specific biomarkers for different types of challenging behaviours among persons with developmental disabilities.

A number of studies have also examined the relationship between parent-report measures of stress, such as the Stress Survey Schedule for Individuals with Autism and other Pervasive Developmental Disabilities (SSS; Groden et al., 2001) or self-reported stress or anxiety, and physiological measures of stress, including cortisol, with mixed outcomes. For example, Corbett and colleagues (2009) examined the link between cortisol and parent-reported stress, measured using the SSS and found that parent-reported stress was negatively correlated with morning cortisol levels but positively correlated with evening cortisol levels. In comparison, an earlier study by the same group (Corbett et al., 2006) did not observe a correlation between cortisol and SSS scores. However, the more extensive cortisol sampling protocol utilised by Corbett et al. (2009), involving the collection of 18 cortisol samples at home over a six day period for each participant rather than the 6 saliva samples collected at home over a two day period by Corbett et al. (2006), may account for the discrepancies between these studies. Lopata and colleagues (2008) demonstrated a positive correlation between self-reported stress and cortisol among children with high-functioning ASDs while Lanni et al. (2012) failed to identify a

correlation between self-reported anxiety and cortisol. A recent study by Bitsika and colleagues (2014) found that among children with ASD, child-reported anxiety was correlated with cortisol levels but that parent-reported anxiety was not. This finding suggests that there may be limitations to parent-report measures of stress for those with ASD, although the communication difficulties associated with ASD may preclude the use of self-report measures for lower functioning individuals. Fewer studies still have examined the relationship between parent-reported stress and challenging behaviour in ASD. One recent study by García-Villamisar and Rojahn (2015) identified a positive correlation between participant's global level of stress, total score on the SSS, and their engagement in repetitive behaviour. Stress was also identified as a unique mediator of the relationship between autistic symptoms and repetitive behaviour.

A greater number of papers have examined LHPA axis functioning in ASD more generally. Corbett and Schupp (2014) examined the cortisol awakening response among 94 male children, 48 of whom were typically developing and 46 who were diagnosed with ASD, and failed to identify any differences between the two groups, a finding which replicates that of Zinke and colleagues (2010). Studies which have examined the diurnal rhythms of cortisol among those with autism, as compared to typically developing controls, have frequently evidenced significantly greater variability in the diurnal cycles of those with ASD than controls (e.g., Corbett et al., 2006; Corbett et al., 2009; Richdale & Prior, 1992). Corbett and colleagues (2009) reported that, among their sample of 44 children, 22 with autism and 22 typically developing controls, participants with ASD presented with a reduced diurnal slope as compared to control participants such that morning cortisol levels were lower and evening cortisol level were higher among participants with ASD than among control participants. Much variability has also been observed across studies which have examined cortisol reactivity to a variety of stressors among participants with ASD (Taylor & Corbett, 2014). Gabriels and colleagues (2013)

have argued that the elucidation of the relationship between challenging behaviours associated with autism and cortisol levels may contribute to our understanding of the variability observed across these studies.

The current study had two key objectives. First, it aimed to compare the cortisol levels of children and adolescents with autism to a sample of age- and gender- matched peers to determine whether differences existed between these groups or between subsets of the ASD sample. Second, it sought to examine the relationship between physiological and behavioural measures of stress and challenging behaviour by assessing the relationship between cortisol and specific topographies of challenging behaviour, specifically SIB, stereotyped or repetitive behaviour, and aggression, among a sample of individuals with ASD.

Experiment 1

Method

Participants. A sample of 104 children and adolescents between the ages of three and 18 years participated in this study. Of these, 52 were independently diagnosed with ASD and 52 were typically developing. Of the typically developing participants, 47 were matched to a participant in the autism group on gender, age (with no more than a 12-month difference between the ages), and pubertal status. The additional five control group participants were matched with a participant in the autism group on age and gender but not pubertal status. Typically developing participants were not diagnosed with a neurodevelopmental disorder, learning disability, or any other psychiatric disorder, nor were they diagnosed with any medical condition or prescribed any medication which may have impacted neuroendocrine functioning. One typically developing participant's cortisol levels fell notably outside of the normal ranges and as a result his data, and the data of the participant with autism with whom he had been matched, were excluded from all analyses. Table 5 provides additional information on the remaining 51 participants in each group.

Participant information was either extracted from participants' records held at their educational placement or sought by contacting parents.

Table 5

Summary of Participant Characteristics of the Autism and Comparison Group for Experiment One

	Autism Group (*n*=51)	**Comparison Group (*n*=51)**
Age	10.97 years (4.06)	10.95 years (4.12)
Gender (male; female)	45; 6	45; 6
Communication Ability	Verbal (*n*=40; 78.4%) Nonverbal (*n*=11; 21.6%)	-
Primary diagnosis	Autism (*n*=45; 88.2%) Asperger's Syndrome (*n*=5; 9.8%) PDD-NOS (*n*=1; 2%)	-
Co-morbid psychiatric diagnoses	None (*n*=13; 25.5%) Intellectual disability (*n*=30; 58.8%) Intellectual disability and ADHD (*n*=2; 3.9%) Attention deficit disorder (*n*=1; 2%) Global developmental delay (*n*=1; 2%) Dyslexia (*n*=3; 5.9%) Dyspraxia (*n*=1; 2%)	-
Co-morbid medical conditions	None (*n*=42; 82.4%) Epilepsy (*n*=5; 9.8%) Partial seizures (*n*=1; 2%) Cerebral palsy (*n*=1; 2%) Epilepsy and Von Willebrand Disease (*n*=1; 2%) Oculofacialcardiodental syndrome (*n*=1; 2%)	-
Psychotropic Medication usage	None (*n*=38; 74.5%) Anticonvulsant (*n*=7; 13.7%) Antipsychotic (*n*=2; 3.9%) Psychostimulant (*n*=2; 3.9%) Antidepressant (*n*=1; 2%) Antipsychotic and Antidepressant (*n*=1; 2%)	-

Note: PDD-NOS=Pervasive Developmental Disorder-Not Otherwise Specified, ADHD= Attention Deficit Hyperactivity Disorder.

Measures.

The Pubertal Development Scale (PDS; Petersen, Crockett, & Richards, 1988).

The PDS was developed as a self-report measure for the assessment of pubertal status, or physical maturation, among children and adolescents. The measure consists of eight items that assess for the physical changes which are associated with puberty. Some items are gender specific, such as those referring to menstruation or breast development for females or the deepening of the voice or growth of facial hair for males. Responses are made on a four-point scale where 1 refers to a physical change that has not begun, and 4 refers to a physical change which appears completed. The reliability and validity of this instrument have been demonstrated (Petersen et al., 1988). In the current study, a parent-report form of the instrument was employed. The decision to use a parent-report version of this measure rather than the self-report version was based on the consideration of the variable level of functioning of participants in the ASD group and the young age of many of the participants. A parent-report form of the instrument has been used and evaluated in previous studies (e.g., Coakley, Holmbeck, Friedman, Greenley, & Thill, 2002; Miller, Tucker, Pasch, & Eccles, 1988; Palmer et al., 2009; Sagrestano, McCormick, Paikoff, & Holmbeck, 1999). A comparison of parent-report, child-report, and doctor-report on the PDS showed a strong correlation between parent-report and child-report for females (r=.75) and males (r=.77) although the level of correspondence between the parent- and child- report differed according to the child's age (Miller et al., 1988).

Saliva collection.

Six samples of saliva were taken from each participant for later cortisol extraction. All saliva samples were collected using a polymer cylindrical swab, the Salimetrics Children's Swab (SCS; Salimetrics Europe, Suffolk, UK). This instrument is 125mm in length allowing it to be held, non-invasively, by the participant or an adult while placed in the mouth. The swab typically collects between 20-100 µl of saliva. As certain foods and

drinks have been found to influence cortisol levels, these were avoided for a minimum of 30 minutes prior to saliva collection. Previous research has shown that the extraction of cortisol from saliva produces similar results to the extraction of cortisol from blood and is a suitable method of cortisol collection in situations where blood sampling would be inappropriate or difficult (e.g., Aardal & Holm, 1995).

Procedure.

The study was approved by an ethical research review board at the National University of Ireland Galway[3]. Consent was sought from parents or caregivers, and participants where possible. For all participants, parents were asked to complete the PDS and to provide demographic information about their child. Following this, samples of salivary cortisol were obtained from each participant over a two-day period. On both days, parents reported their child's time of awakening in order to ensure that the cortisol awakening response had passed prior to the collection of the first saliva sample. Saliva samples were collected at 10 a.m., 12.30 p.m., and 2.30 p.m, a cortisol collection schedule previously employed by Devlin, Healy, Leader, & Hughes (2011), at the children's school. During data collection, the SCS was placed in participants' mouths, under the tongue when tolerated, for 90 seconds, timed using a digital timer, at each saliva collection timepoint. Following this, the swab was placed in a plastic vial, labeled with the participant's ID code, the date, and time of sample collection. The swabs were held using the protective plastic covering supplied which ensured that all saturated swabs were not handled or contaminated. All saliva samples were refrigerated immediately, and subsequently frozen at -20^{O}C until assayed for cortisol levels.

[3] The first year of this research program was conducted at the National University of Ireland, Galway. Ethical approval was awarded by NUI Galway's research ethics committee during this year and this was accepted by Trinity College Dublin as sufficient evaluation of the ethical considerations of the research program.

Cortisol analysis.

Saliva samples were assayed for cortisol levels in the third author's laboratory at the National University of Ireland, Galway. Prior to assaying, all samples were thawed, and centrifuged at 3000rpm for 15 minutes. Samples were assayed in duplicate using a commercial available salivary cortisol enzyme immunoassay kit (Salimetrics Europe, Suffolk, UK) over 4 days in accordance with manufacturer's instructions. The sensitivity of this assay ranged from 0.003-3.0 micrograms per decilitre (μg/dl). The co-efficient of variability, a measure of the precision or accuracy of the assay, was assessed. The intra-assay coefficient of variability was found to be 7.1%, while the inter-assay coefficient of variability was found to be 8.9%. Salimetrics (n.d.) proposes that intra-assay coefficient of variability should be below 10% while inter-assay coefficients of variability below 15% demonstrate high reproducibility and precision in the assay.

Statistical Analysis.

Inspection of the data revealed few missing cortisol data points (1.5%). Among participants with ASD, there were five measurement timepoints (1.6%) for which cortisol values were missing due to either the non-collection of a saliva sample at that timepoint or the collection of an insufficient volume of saliva for analysis purposes. Among control participants, there were a total of four (1.3%) measurement timepoints for which cortisol data was missing. These missing data points did not impact upon analyses as similar to Gabriels et al. (2013), the cortisol values at each measurement timepoint were averaged across the two days of sampling to reduce any potential impact of outliers, and at least one cortisol sample was available for all participants at each of the cortisol measurement timepoints.

Cortisol data were found to be positively skewed, as is typical with cortisol distributions (Adam & Kumari, 2009). As a result, a natural logarithmic transformation was applied in order to normalise the distribution of the data. However, as analyses

revealed that the use of the log-transformed data did not produce statistical outcomes different to the use of the non-transformed data, all subsequent analyses were performed, and reported, using the non-transformed data.

In order to compare whether a diagnosis of autism impacted upon cortisol levels at any of the three measurement timepoints, two 2x3 repeated measures ANOVAs were employed. For both, the within-subjects factors were diagnosis with two levels (autism or typically developing) and cortisol measurement time with three levels (sample one, sample two, sample three), and the dependent variable was cortisol values. The first analysis assessed for differences in cortisol between the pairs matched on age and gender only (n=51). The second analysis assessed for differences between the pairs matched on age, gender, and pubertal status (n=46). Following this, a series of 2x3 mixed ANOVAs were used to examine differences between various subsets of the autism group sample across the repeated cortisol measurements. For these analyses, the between-subject factor was the factor differentiating the groups (e.g., presence or absence of co-morbid intellectual disability, medication status, verbal capability, or age), with two levels in each case, the within-subjects factor was cortisol measurement time (sample one, sample two, sample three), and the dependent variable was cortisol value. The groups compared were: (1) Participants with ASD and a co-morbid intellectual disability and participants with ASD and no co-morbid intellectual disability; (2) Participants with ASD who were medicated and participants with ASD who were unmedicated; (3) Participants with ASD who were verbal and participants with ASD who were nonverbal, and (4) Children with ASD (3-11 years) and adolescents with ASD (12-18 years).

Finally, the coefficient of variation was calculated to provide a measurement of variability in cortisol levels for each participant. A dependent samples t-test was subsequently used to assess for significant differences in cortisol variability between the two groups. All data were analysed using SPSS version 21.

Results and Discussion

Descriptive data were initially calculated to characterise the mean cortisol values at each sample for each group. For the ASD group, mean cortisol values at sample one were found to be 0.25 μg/dl (*SD*=0.16 μg/dl), 0.20 μg/dl (*SD*=0.16 μg/dl) at sample two, and 0.17 μg/dl (*SD*=0.10) at sample three. For typically developing participants, cortisol values at sample one were found to be 0.19 μg/dl (*SD*=0.10 μg/dl), 0.17 μg/dl at sample two (*SD*=0.07), and 0.17 μg/dl (SD=0.11 μg/dl). These values are represented graphically in Figure 2 which indicates that the autism group appeared to have higher cortisol values at sample one and sample two. The first of the 2x3 repeated measures ANOVAs assessed whether this difference was statistically significant between participants with autism and controls matched on age and gender. Mauchly's test of sphericity was not significant for any of the variables (all *p*'s>.05) indicating that the assumption of sphericity had been met. A main effect of cortisol measurement time was observed, $F(2,100)=8.27$, $p<.001$. Contrasts revealed that cortisol levels at sample one were significantly higher than cortisol levels at sample three. Neither a significant main effect of diagnosis, $F(1,50)=2.95$, $p=.09$, $d=0.34$, nor a significant interaction between diagnosis and cortisol measurement time, $F(2,100)=2.73$, $p=.07$, $d=0.33$, was observed. The second 2x3 repeated measures ANOVA, assessing for differences between pairs matched on age, gender, and pubertal status, produced similar outcomes. Mauchly's test of sphericity revealed that the assumption of sphericity had been met. Once again, a main effect of cortisol measurement time was observed, $F(2, 90)=6.34$, $p=.003$, with contrasts revealing that the significant differences were attributable to a significant difference between cortisol measure values at samples one and three as reflected in Figure 2. As in the previous analysis, there was no evidence for a significant main effect of diagnosis, $F(1,45)=2.33$, $p=.13$, $d=0.31$, nor was there a significant interaction between diagnosis and cortisol measurement time observed, $F(2,90)=2.47$, $p=.09$, $d=0.33$.

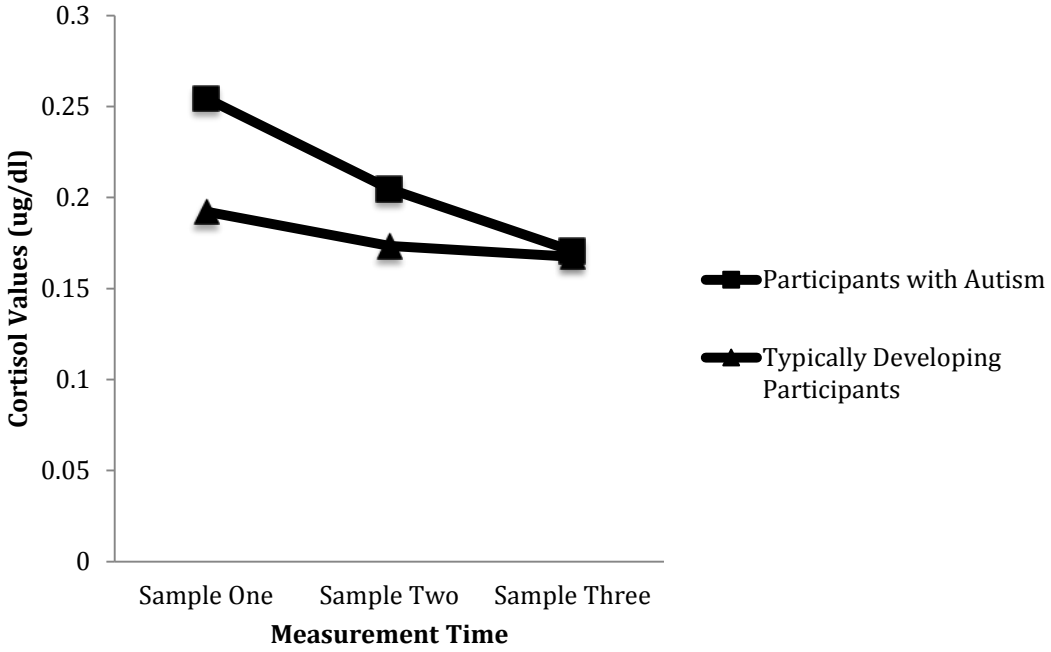

Figure 2. Mean cortisol values at each measurement time for both groups.

For each of the four 2x3 mixed ANOVAs, a main effect of time of cortisol measurement was observed suggesting that cortisol values differed depending on the time of measurement as would be expected given the established diurnal rhythms of cortisol (Adam & Kumari, 2009). For the first comparison, participants with ASD and a co-occurring intellectual disability (n=32) and participants with ASD and no co-occurring intellectual disability (n=19), there was no significant main effect of the presence or absence of a co-occurring intellectual disability identified, $F(1,49)=.01$ $p=.92$, $d=0.03$, nor was there a significant interaction between the presence or absence of a co-occurring intellectual disability and time of cortisol measurement discernible, $F(2,98)=1.55$, $p=.22$, $d=0.36$. For the second comparison, participants with ASD who were taking psychotropic medications (n=13) and unmedicated participants with ASD (n=38), there was no significant main effect of medication, $F(1,49)=.17$, $p=.68$, $d=0.13$, nor was there a significant interaction between medication status and time of cortisol measurement,

$F(2,98)=.97$, $p=.38$, $d=0.32$. The third comparison, participants with ASD who were verbal ($n=40$) and those who were nonverbal ($n=11$), revealed no significant main effect of verbal capability, $F(1,49)=.00$, $p=.99$, nor a significant interaction between verbal capability and time of cortisol measurement, $F(2,98)=.02$, $p=.98$, $d=0.05$. The final comparison, children with ASD ($n=32$) and teenagers with ASD ($n=19$), also failed to reveal a significant main effect of age, $F(1,49)=.79$, $p=.38$, $d=0.26$ or a significant interaction between age and time of cortisol measurement, $F(2,98)=.27$, $p=.76$, $d=0.15$.

While the mean coefficient of variation was higher in the autism group ($M=0.39$, $SD=0.20$) than in the typically developing control group ($M=0.36$, $SD=0.25$), a paired-samples t-test indicated that this difference was not statistically significant, $t(50)=.75$, $p=.46$, $d=0.15$.

The findings from Experiment 1 indicated no significant difference in the cortisol levels of children and adolescents with ASD and their typically developing peers. However, Cohen's *d* indicated that small main effects and interaction effects were observed. It was not possible to compare the size of these effects with previous studies which have identified significant differences in diurnal cortisol (e.g., Corbett et al., 2009) or those which have reported non-significant differences in diurnal cortisol (e.g., Corbett et al., 2006) between participants with autism and typically developing participants as these studies do not provide information on effect sizes observed. However, future research employing a larger sample size may further our understanding of the nature of any differences in diurnal cortisol existing between individuals with autism and typically developing persons. Both groups showed the expected decline in cortisol levels across the day (consistent with typical diurnal pattern), and evinced similar degrees of variability. Results also suggest that the presence or absence of a co-occurring intellectual disability, use of psychotropic medication, verbal capability, and age were unrelated to cortisol levels among the participants with ASD.

Experiment 2

Method

Recruitment and Participants. Participants in both experiments of this research study were recruited using convenience sampling. A variety of recruitment methods were used including: direct contact with special schools or ASD-specific classrooms via telephone, online postings on various Irish social media pages and groups (e.g., Irish Autism Action, ABA Ireland Facebook, Rollercoaster.ie etc.) frequented by the parents or educators of children with developmental disabilities; direct contact, via phone or email, with behaviour analysts working in services for persons with developmental disabilities; and the creation of posters advertising the study which were sent to schools known to contain classrooms specific to pupils diagnosed with ASD.

In total, sixty-one children and adolescents independently diagnosed with an ASD participated in this study. Of these, 51 had previously participated in Experiment 1. An additional six participants were recruited but not included in the final sample as they either failed to comply with cortisol collection procedures by blocking the swab from entering their mouth or refusing to allow the swab to remain in their mouth for a sufficient period of time (n=2), had a co-morbid behavioural or psychiatric symptom (i.e., severe aggressive behaviours, severe food refusal, or depressive episode) that made their participation in the research study inappropriate at that time (n=3), or indicated verbally that they did not wish to participate when approached following the acquisition of parental consent (n=1). Due to a paucity of cortisol collection materials, it was not possible to habituate the two participants who presented with issues relating to compliance with cortisol collection to the procedure. As a result, additional research participants were recruited in their stead. Table 6 outlines the characteristics of this sample. As in Experiment 1, participant information was compiled through record review and parent-survey.

Table 6

Summary of Participant Characteristics for Experiment Two

	Participant Characteristics
Age	10.89 years (4.25)
Gender (male; female)	53; 8
Communication Ability	Verbal (n=49; 80.3%) Nonverbal (n=12; 19.7%)
Primary diagnosis	Autism (*n*=55; 90.2%) Asperger's Syndrome (*n*=5; 8.2%) PDD-NOS (*n*=1; 1.6%)
Co-morbid psychiatric diagnoses	None (*n*=15; 24.6%) Intellectual disability (*n*=37; 60.7%) Intellectual disability and ADHD (*n*=3; 4.9%) Dyslexia (*n*=3; 4.9%) Attention deficit disorder (*n*=1; 1.6%) Global developmental delay (*n*=1; 1.6%) Dyspraxia (*n*=1; 1.6%)
Co-morbid medical conditions	None (*n*=50; 82%) Epilepsy (*n*=5; 8.2%) Partial seizures (*n*=1; 1.6%) Cerebral palsy (*n*=1; 1.6%) Hydrocephalus (n=1; 1.6%) Epilepsy and Von Willebrand Disease (*n*=1; 1.6%) Klinefelter syndrome (*n*=1; 1.6%) Oculofacialcardiodental syndrome (*n*=1, 1.6%)
Psychotropic Medication usage	None (*n*=46; 75.4%) Anticonvulsant (*n*=7; 11.5%) Psychostimulant (*n*=3; 4.9%) Antipsychotic (*n*=2; 3.3%) Antidepressant (*n*=1; 1.6%) Antipsychotic and Antidepressant (n=1; 1.6%) Antipsychotic, Psychostimulant, and Anti-convulsant (n=1; 1.6%)

Note: PDD-NOS= Pervasive Developmental Disorder-Not Otherwise Specified, ADHD=Attention Deficit Hyperactivity Disorder.

The Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003).

The SCQ is a 40-item parent-report measure that assesses for the presence of behaviours that are characteristic of autism. This measure was developed from the Autism Diagnostic Interview- Revised (Lord, Rutter, Le Couteur, 1994). Items are scored either 0, symptom

not present, or 1, symptom present. Scores may range from 0-39 with higher scores indicative of more prevalent symptoms of autism and a greater severity.

The Stress Survey Schedule for Individuals with Autism and other Developmental Disabilities (SSS; Groden et al., 2001). The SSS is a 49-item instrument is designed to assess stress among persons with ASD. It consists of eight subscales which evaluate participants' reactions to different categories of stressors which are prevalent in ASD. These subscales are: Anticipation/Uncertainty, Changes and Threats, Unpleasant Events, Pleasant Events, Sensory/Personal Contact, Food Related Activity, Social/Environmental Interactions, and Ritual Related Stress. The psychometric properties, including validity and internal consistency, of the instrument have been demonstrated (Groden et al., 2001; Goodwin, Groden, Velicer, & Diller, 2007).

The Repetitive Behaviors Scale- revised (RBS-R; Bodfish, Symons, & Lewis, 1999; Bodfish, et al., 2000). The RBS-R is a rating scale designed to assess the frequency or severity of repetitive, stereotyped, behaviours. This measure was employed and scored as per the alternative method proposed by Lam (2004). In this way, it consists of five subscales which assess distinct forms of repetitive behaviour. These subscales are: Stereotypic Behavior, Self-Injurious Behavior, Compulsive Behavior, Ritualistic/Sameness Behavior, and Restricted Interests. The reliability and validity of this instrument for use with individuals diagnosed with ASD have been assessed and found to be adequate (Bodfish et al., 2000).

The Behavior Problems Inventory (BPI-01; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001). The BPI-01 is a 52-item respondent-based rating scale which is designed to assess the frequency and severity of challenging behaviours, specifically SIB, stereotypic behaviours, and aggressive/destructive behaviours, of individuals diagnosed with an intellectual disability or other developmental disability. Several studies have found the BPI-01 to be a valid and reliable measure of challenging behaviour among persons with

developmental disabilities (Gonzalez et al., 2009; Rojahn et al., 2001; Rojahn, Wilkins, Matson, & Boisjoli, 2010).

Saliva collection. The saliva collection procedure was identical to that described in Experiment 1.

Procedure. Ethical approval for this study was obtained from the research ethics committee at the National University of Ireland, Galway. Following this, consent was sought from parents or caregivers, and participants where possible. All parents were asked to complete a questionnaire pack containing the PDS, the SCQ, and the SSS. For each participant, a teacher or their key instructor was also asked to fill out the RBS and the BPI-01 in order to obtain information on any challenging behaviours with which participants presented. Following this, samples of salivary cortisol were obtained from each participant over a two-day period, as in Experiment 1.

Cortisol analysis. The cortisol analysis procedures were the same as those described in Experiment 1. For Experiment 2, the intra-assay coefficient of variability was found to be 6.86%, while the inter-assay coefficient of variability was found to be 8.9%.

Statistical analysis. Data were missing for a number of variables including: cortisol data (2.19%), SCQ items (2.07%), SSS items (5.42%), RBS-R items (2.7%), and BPI-01 items (3.97%). The impact of missing data on scale and subscale scoring was dealt with by using the average score per item from the available items as the subscale score. Total scale score was then computed by calculating the average subscale score.

Initially, we examined the correlations between participants' physiological stress (i.e., average daily cortisol level), parent-reported stress (i.e., SSS total score), and subscale and total scores on the measures of challenging behaviour (i.e., RBS-R and BPI-01)

Next, a series of hierarchical multiple regressions was conducted to examine whether parent-reported stress (i.e., SSS total score) or physiological stress (i.e., average

daily cortisol level) were predictive of the frequency or severity of challenging behaviour (i.e., Stereotypy Frequency (BPI-01), Stereotypy Severity (BPI-01), Self-injurious behaviour Frequency (BPI-01), Self-injurious behaviour Severity (BPI-01), Aggression Frequency (BPI-01), Aggression Severity (BPI-01), and Stereotypy Severity (RBS-R)). For each regression analysis, both stress variables were first entered as predictors. Following this, the impact of controlling for age and SCQ score on the statistical outcome of each regression was examined. For all regression analyses conducted, the number of predictor variables entered did not exceed the criterion of $n \geq 20+5k$ suggested by Khamis and Kepler (2010).

Following this, for each topography of challenging behaviour, participants falling within the upper quartile of scores on that parent-report subscale or measure (i.e., stereotyped, self-injurious, or aggressive/destructive behaviour subscales of the BPI-01, and RBS-R total score) were categorised as "high", and participants falling within the lower quartile of scores were categorised as "low". The average daily cortisol level and parent-reported stress of the high and low challenging behaviour groups were compared using an independent samples *t*-test. A Bonferroni correction of .05/8=.008 was used to adjust the alpha level to control for the use of multiple univariate analyses. These analyses were conducted in order to allow for a close comparison between our results and those of previous similar research studies (Symons et al., 2003; Symons et al., 2011; Gabriels et al., 2013) that recruited participants on the basis of their engagement in either high or low levels of specific challenging behaviours. For example, Symons et al. (2003) recruited participants engaging in high or low levels of SIB measured using the BPI-01, while Gabriels et al. (2013) recruited participants engaging in high or low levels of repetitive behaviour based upon their RBS-R scores. Thus, stratifying our sample in this manner allowed us to compare our results with the findings of these studies although our recruitment processes differed.

Next, we examined the correlation between parent-reported stress and average daily cortisol level. Finally, participants falling within the upper quartile of scores on the SSS were categorised as a “high stress” group and those falling within the lower quartile of scores on the SSS were categorised as a “low stress” group. The average daily cortisol level of these two groups was compared using an independent samples *t*-test.

Results and Discussion

Table 7 presents the Pearson Product Moment correlations between parent-reported stress (i.e., SSS total score), physiological stress (i.e., average daily cortisol level), and measures of challenging behaviour (i.e., total scores and subscale scores of the BPI-01 and the RBS-R). No significant correlations between stress, parent-reported or physiological, were identified (see Table 7).

Table 7

Pearson Product Moment Correlations between Parent-reported Stress, Physiological Stress, and Measures of Challenging Behaviour

	Correlation Coefficients	
	Parent-Reported Stress (SSS Total Score)	**Physiological Stress (Average Daily Cortisol Level)**
Self-injurious Behaviour Frequency (BPI-01)	-.11	-.02
Self-injurious Behaviour Severity (BPI-01)	-.11	-.03
Stereotyped Behaviour Frequency (BPI-01)	.07	.12
Stereotyped Behaviour Severity (BPI-01)	.05	.08
Aggressive/Destructive Behaviour Frequency (BPI-01)	.17	.02
Aggressive/Destructive Behaviour Severity (BPI-01)	.07	-.01
Stereotypic Behaviour (RBS-R)	-.16	.01
Self-injurious Behaviour (RBS-R)	-.20	-.01
Compulsive Behaviour (RBS-R)	.06	.04
Ritualistic/Sameness Behaviour (RBS-R)	.23	-.13
Restricted Interests (RBS-R)	-.10	-.04
RBS-R Total Score	-.06	-.05

Note: None of the correlations presented in this table achieved statistical significance.

A series of regression analyses were also conducted (see Table 8). Results revealed that a model comprising the two indices of stress (i.e., parent-reported stress, average cortisol level) was not a significant predictor of any of the challenging behaviour variables. It was also demonstrated that, for each regression, accounting for age or SCQ score in Step One, and then exploring the relationship between stress and challenging behaviour, did not impact upon the pattern of results (in the interest of brevity these analyses are not reported here).

Table 8

Outcomes of Multiple Regression Analyses for BPI-01 Frequency and Severity Subscale Scores, and RBS-R Total Score

Variable	*b*	*SE b*	β	ΔR^2
Stereotypy Frequency (BPI-01)				
Predictor: Parent-reported stress (SSS total)	.076	.115	.090	-.016
Predictor: Average cortisol level	.798	.875	.123	-.016
Stereotypy Severity (BPI-01)				
Predictor: Parent-reported stress (SSS total)	.030	.071	.057	-.028
Predictor: Average cortisol level	.377	.618	.083	-.028
Self-injurious behaviour Frequency (BPI-01)				
Predictor: Parent-reported stress (SSS total)	-.041	.048	-.114	-.022
Predictor: Average cortisol level	-.097	.365	-.036	-.022
Self-injurious behaviour Severity (BPI-01)				
Predictor: Parent-reported stress (SSS total)	-.035	.039	-.120	-.020
Predictor: Average cortisol level	-.102	.294	-.046	-.020
Aggressive/destructive behaviour Frequency (BPI-01)				
Predictor: Parent-reported stress (SSS total)	.109	.082	.177	-.033
Predictor: Average cortisol level	.290	.626	.062	-.003
Aggressive/destructive behaviour Severity (BPI-01)				
Predictor: Parent-reported stress (SSS total)	.039	.074	.072	-.031
Predictor: Average cortisol level	.099	.637	.021	-.031
Repetitive Behaviour (RBS-R)				
Predictor: Parent-reported stress (SSS total)	-.032	.067	-.064	-.031
Predictor: Average cortisol level	-.142	.507	-0.38	-.031

These results suggest that stress, as reported by parents and measured physiologically, was not correlated with or predictive of the severity or frequency of SIB, aggression, and stereotyped behaviour among the current sample of children and adolescents diagnosed with autism.

Comparisons of those presenting with high (n=18) or low (n=23) levels of SIB revealed no significant differences in mean daily cortisol, $t(39)=-.49$, $p=.63$ nor in parent-reported stress, $t(38)=-.21$, $p=.83$, between the two groups. The comparison of those presenting with high (n=16) and low (n=20) frequencies of aggressive/destructive behaviour also revealed no differences in mean daily cortisol level, $t(34)=-.05$, $p=.96$, nor

in parent-reported stress, $t(33)=.50$, $p=.62$, between these two groups. There was also no difference between those categorised as high on the RBS-R ($n=15$) and those receiving low scores ($n=14$) on this measure in mean daily cortisol levels, $t(27)=-.59$, $p=.56$, nor in parent-reported stress, $t(26)=-.27$, $p=.79$. However, those scoring high on the stereotyped behaviour frequency subscale of the BPI-01 ($n=18$) had higher cortisol levels ($M=0.21$, $SD=0.07$) than those receiving low scores on this measure ($n=18$; $M=0.16$, $SD=0.04$), but although a large effect size was observed this difference was statistically non-significant, $t(28.27)=2.5$, $p=.02$, $d=0.88$, following the application of the Bonferroni correction ($p<.008$).

As the Stereotyped Behaviour subscale of the BPI-01 assesses stereotyped motoric behaviour, and the RBS-R total score is computed from participants' scores on a variety of subscales (assessing repetitive SIB, compulsive behaviour, ritualistic/sameness behaviour, and restricted interests), we compared those who had scored in the upper ($n=15$) and lower ($n=16$) quartiles of the stereotypic behaviour subscale of the RBS-R only. Results showed no significant differences between these groups in average daily cortisol levels, $t(29)=.05$, $p=.96$, or in parent-reported stress levels, $t(28)=-1.17$, $p=.25$.

We also examined the correlation between parent-reported stress and average daily cortisol level among the participants with autism. This analysis failed to reveal any discernible relationship between the two variables, $r(59)=-.16$, $p=.23$. Finally, we compared the cortisol levels of the "high stress" group ($n=15$) and the cortisol levels of the "low stress" group ($n=15$). No significant differences in cortisol between these groups were found, $t(28)=-1.029$, $p=.32$.

The results reported here are in contrast with previous findings in this area (e.g., Gabriels et al., 2013; Symons et al., 2003; Symons et al., 2011; Verhoeven et al., 1999). Cortisol levels reported in the current study did not differ based on the presence of SIB. In addition, there was no apparent association between engagement in high or low levels of

aggressive/destructive behaviour and levels of cortisol. Furthermore, the direction of the relation between cortisol levels and engagement in stereotypy was in contrast to findings reported by Gabriels and colleagues (2013). The results reported here tentatively suggest that those engaging in a high frequency of stereotypy, measured by the BPI-01, may present with higher cortisol levels than those engaging in little or no motor stereotypy. While the difference between these groups was statistically non-significant following the application of a Bonferroni correction, the large effect size (d=0.88) observed may suggest that this relation may have been significant with a larger sample size.

General Discussion

The current study aimed to examine whether cortisol levels, or variability in cortisol levels, differed among participants with ASD and a sample of matched typically developing controls. It also sought to explore the relationship between physiological and parent-report measures of stress and various topographies of challenging behaviour among a sample of 61 children and adolescents diagnosed with ASD.

A key finding emerging from this research was that those with ASD engaging in the highest frequencies of stereotyped behaviour measured by the BPI-01 presented with higher cortisol levels, although this difference was statistically non-significant, than those with ASD who engaged in little or no stereotyped behaviour, although no correlation between these variables was discernible for the whole sample. Cohen's *d* indicated that this was a large effect, with the average cortisol value for the high stereotypy group found to be 23.8% higher than that of the low stereotypy group. The average daily cortisol level of the typically developing participants (M= 0.18; SD=0.07) fell midway between the average daily cortisol levels of the low stereotypy group (M=0.16, SD=0.04) and the high stereotypy group (M=0.21, SD=0.07). While these results tentatively support suggestions of a relationship between stereotypy and stress, the direction of the association observed conflicts with that of a recent study reported by Gabriels and colleagues (2013). The

authors identified significantly lower cortisol levels among a high stereotypy group than among a low stereotypy group, with a difference of 36% in the cortisol levels of each group. There were a number of differences between the current study and that of Gabriels et al. (2013) which may have contributed to the differences in outcomes observed. Gabriels and colleagues recruited participants on the basis of their high or low score on the RBS-R, only included pre-pubescent males with an IQ of greater than 40, used filter papers to collect saliva samples, and used a different time schedule for saliva sample collection than that employed in the current study. Future research is necessary to determine whether any of these specific variables may impact on the outcomes of studies such as those reported here.

The current findings appear to suggest that stereotypy may be an overt manifestation of stress among individuals with ASD but that it does not appear to function in a way that mitigates this stress. Many researchers have previously noted that stereotyped behaviours are often associated with stress or stressful situations (Hutt & Hutt, 1968; Militerni, Bravaccio, Falco, Fico, & Palermo, 2002; Schlagger & Mink, 2003), and are frequently considered behavioural signs of stress among animals (e.g., Broom & Johnson, 1993; Morgan, 2006). Groden et al. (1994) also theorised that stereotyped behaviours may represent faulty coping mechanisms to stressors among those with autism. Given the discrepant findings between this study and that of Gabriels et al. (2013), further studies are required to investigate the relationship between stress, cortisol, and repetitive behaviour in greater detail.

The current study did not identify a relationship between cortisol levels and SIB and this finding is also in contrast with previous research in this area (e.g., Symons et al., 2003; Symons et al., 2011; Verhoeven et al., 1999). Verhoeven et al. (1999) identified lower cortisol levels among those who engaged in frequent SIB whilst Symons et al. (2003; 2011) identified higher cortisol levels among those who engaged in frequent SIB.

The inconsistency in findings across these and the current study may be attributable to differences in the samples employed. Previous studies in the areas have used samples of adults with various developmental disabilities selected on the basis of their severe or chronic SIB (e.g., Symons et al., 2003; Symons et al., 2011). However, the current study examined the relationship between cortisol and self-injury among a sample of children (3-18 years) with autism as a primary diagnosis who were recruited using convenience sampling rather than selected on the basis of their engagement in particular challenging behaviours.

To our knowledge, previous research has not examined the relationship between cortisol levels and aggression among individuals with developmental disabilities, thus a comparison cannot be made between our findings and any others. However, there is research to suggest that a relationship may exist between stress and aggression for persons with developmental disabilities. Moskowitz and colleagues (2013) found evidence that the challenging behaviours, including aggressive behaviours, of three children diagnosed with ASD occurred more frequently during high-anxiety contexts than during low-anxiety contexts. The authors suggested that the elevated physiological arousal produced by, and stress associated with, high-anxiety contexts may have contributed to participants' engagement in challenging behaviours. Relatedly, To and Chan (2000) found that progressive muscle relaxation training, intended to reduce stress and anxiety, led to reductions in the aggressive behaviour of a sample of individuals with intellectual disabilities. Thus, while the findings of the current study suggest little correspondence between stress and SIB and aggression, future research that examines this relationship among a sample of individuals selected on the basis of their engagement in severe aggressive/destructive behaviour may produce different outcomes.

The lack of a relationship between parent-reported stress and physiological stress for the frequency and severity of challenging behaviour among the participants with ASD

within the current study is also of note. A connection between these variables has been theorised (e.g., Groden et al., 1994) but infrequently empirically assessed. The results of the current study suggest that the association between these variables is not uniform among all individuals with ASD, but that it may exist for some individuals (i.e., those evincing high levels of stereotyped behaviours). Future research should examine whether behavioural function impacts upon the relation between physiological activity and challenging behaviour. It is possible that behaviours which serve a social function (socially mediated) may be unrelated to internal physiological states but that those which are automatically reinforced (independent of the social environment), as stereotyped behaviour most commonly is (e.g., Hanley et al., 2003; Healy, Brett & Leader, 2013), may be correlated with physiological activation.

The lack of a correlation between parent-reported stress on the SSS and cortisol levels observed in the current study is also of interest. Previous studies in this area have reported various findings with some studies identifying a correlation between the two variables (e.g., Corbett et al., 2009) and others failing to demonstrate any relationship or correlation (e.g., Corbett et al., 2006). Bitsika and colleagues (2014) suggested that parent-report measures may not accurately capture a child's experience of anxiety, while children's self-reported anxiety may be more strongly linked to physiological activity. Previous research has suggested that a desynchrony may exist between physiological activity and overt behaviour among individuals with ASD (see Lydon et al., in press/Study 1: Chapter 2) and this may explain the lack of predictive validity of parent ratings of stress inferred from the observation of overt behaviours. This finding suggests that reliance on behavioural observation as the sole manner of understanding the experiences of individuals with developmental disabilities should be cautiously considered, and that the more frequent use of physiological recording in order to gain an insight into the emotional states of individuals with autism and other developmental disabilities may be recommended.

In the current sample, both typically developing participants and participants with autism demonstrated cortisol levels consistent with typical diurnal patterns across the measurement timepoints (i.e., highest mean cortisol level at the morning sample followed by a decrease in cortisol across the subsequent two measurement timepoints). A small but non-significant effect of autism diagnosis on stress was observed whereby participants with autism may have had higher stress than their typically developing peers. Previous research in this area which has yielded discrepant findings may be related to the size of diurnal cortisol differences between these populations and differing sample sizes. However, a review by Taylor and Corbett (2014) suggested that differences in cortisol secretion and diurnal rhythms may be confined to those individuals with ASD who are low-functioning and may not emerge in the comparison of high-functioning individuals with ASD and their typically developing peers. The current study employed a diverse sample across the spectrum, with some participants diagnosed with severe intellectual disabilities and others presenting with no cognitive delays and functioning within mainstream settings. Therefore, it may be possible that significant differences in diurnal cortisol would have been observed with a more homogenous sample of participants with ASD. While our analysis did not suggest a difference in cortisol levels based on the presence or absence of a co-occurring intellectual disability, it may be that the level of functioning of these individuals was insufficiently characterised by simply viewing it in terms of a co-occurring diagnosis intellectual disability rather than precise estimates of IQ or verbal ability. The validity of the results obtained, however, is underscored by the use of age, gender-, and pubertal status-matched controls. The lack of differences in the variability of cortisol levels between the typically developing and autism group was somewhat surprising. Taylor and Corbett (2014) found that previous research is suggestive of greater intra- and inter-individual variability among persons with ASD. However, it is possible that our results were influenced by our measurement of cortisol levels during the

morning and early afternoon, and our non-measurement of the cortisol awakening response or of evening cortisol levels. It is possible that between-group differences in cortisol levels or variability may have emerged in cortisol measures taken at these times.

Non-measurement of the cortisol awakening response or evening cortisol levels may be considered a criticism of this study. However, the circadian rhythms of cortisol among persons with autism have been examined elsewhere in methodologically strong studies (e.g., Corbett et al., 2009; Corbett & Schupp, 2014). Given that the aim of the current study was foremost to examine the relationship between stress and challenging behaviour, following the demonstration that levels of physiological stress were comparable among participants with autism and typically developing peers, a more extensive cortisol measurement protocol was deemed unnecessary.

The current study had a number of limitations. The participant characterisation of the current study may be considered a limitation. We relied upon, and reported herein, the results of independent psychological assessments for participants with ASD taken from extant child records. In this way, participants' diagnoses were not verified by the researchers. This research also relied upon convenience sampling and it may be argued that different results would have been obtained had individuals with ASD and specific behavioural profiles (e.g., high/low rates of stereotypy, SIB and aggression) been recruited. Also, while the comparison of typically developing participants and participants with ASD in Experiment 1 did not yield statistically significant differences, Cohen's *d* suggested that small effects were observed. In this way, it is possible that these analyses were underpowered, although our sample size exceeded that of previous similar research studies (e.g., Corbett et al., 2006; Corbett et al., 2009). Future research studies employing larger sample sizes may further our understanding of any abnormalities in the diurnal cortisol levels of persons with ASD.

An additional limitation of the current study was the potential mismatch between the timescales of the independent and dependent variables. Cortisol can be best conceptualised as a state measure, measured three times daily over two days, while the measures of challenging behaviour are akin to trait measurement given their assessment of behavioural tendencies over a longer period of time. This may be considered a methodological flaw of the current study and others reporting similar measures (e.g., Corbett et al., 2006; Corbett et al., 2009; Gabriels et al., 2013) of the relation between psychological variables (e.g., anxiety, stress, worry) assessed over an extensive period of time and specific time-point measures of physiological activity. This possible measurement discrepancy may contribute to the explanation of our findings regarding the association between cortisol and stereotyped behaviour. Participants classified as engaging in a "low" frequency of stereotypy were those who were reported to engage in stereotyped motor behaviour either never or monthly as per the BPI-01 rating scale, while those engaging in a "high" frequency of challenging behaviour were reported to engage in the behaviour hourly. In this way, the measures of challenging behaviour were likely more sensitive for these participants regarding the presence or absence of stereotyped behaviour proximal to the cortisol measurement than for the remainder of the sample who engaged in stereotypy either weekly or hourly. This explanation is plausible; other researchers (e.g., Sonnenschein et al., 2007) have found that the detection of variables relating to cortisol activity is more likely with ambulatory assessment than with measures requiring retrospective recall. Future research examining the relationship between cortisol and challenging behaviour may benefit from utilising the more objective techniques of behavioural observation (i.e., measurement of stereotypy frequency/duration) during the same time periods as the cortisol measurement.

The current research also had a number of strengths including the diversity of the sample recruited, which included individuals at varying ranges of severity. There has been

a predominant focus on high-functioning individuals in studies of physiological activity in ASD (see Lydon et al., in press/Study 1: Chapter 2). In this way, the demonstration of the feasibility of collecting measures of cortisol from low-functioning individuals is important; only six participants were recruited who were either unable or unwilling to participate, while most of those who successfully participated were diagnosed with co-occurring intellectual disabilities. Further, the matching of participants in the experimental and control groups on age, gender, and pubertal status may be considered a strength of the study and contributes to its internal validity. Similarly, the examination of the impact of psychotropic medication usage, pubertal status, age, and severity of impairment (i.e., presence or absence of a co-occurring intellectual disability) on cortisol levels may also be considered a strength as the failure to account for these variables by other studies examining cortisol in ASD has been noted (Taylor & Corbett, 2014).

Future research should attempt to further examine and characterise those who present with high or low levels of stereotypy in order to determine additional differences between these groups that may potentially mediate the conflicting findings between stereotypy and stress observed here and in Gabriels et al. (2013). For example, stereotyped motor behaviour is more commonly observed among lower-functioning individuals with ASD (Bodfish et al., 2000; Goldman et al., 2009; Healy & Leader, 2011) than among their higher-functioning counterparts. Although our results did not suggest that the presence or absence of an intellectual disability, or verbal capability, resulted in differences in cortisol levels, it is possible that differences may emerge in a carefully characterised sample of participants with ASD. It was not feasible to assess IQ and language ability in the current study, and thus we relied upon extant independently conducted psychological assessments and parent-report of verbal capability. However, the large effect size of the non-significant findings in this study (d=0.88), and of the findings of Gabriels et al. (2013), suggests that future research could explore the relationship between stereotypy and stress by carefully

categorising participants on the basis of their engagement in high or low levels of stereotypy, for whom administration of measures of intellectual and verbal functioning may be more feasible.

It is certainly plausible that individual characteristics may result in differential physiological activity among individuals with ASD. Several studies have found evidence of such moderation of physiological activity by a number of participant variables. Level of cognitive functioning has been implicated as potentially impacting upon physiological activity by a recent review of studies examining cortisol in autism (Taylor & Corbett, 2014). This suggests that physiological dysregulation in cortisol rhythms may exist only among individuals with autism who are lower functioning and not among their higher-functioning counterparts. Kootz and colleagues (1982) also noted differences in physiological activity between participants with autism who were either low or high functioning during exposure to a variety of stimuli and learning tasks. Language ability has also been suggested to impact upon physiological activity. A recent study by Stagg et al. (2013) found that individuals with autism and no language delay had similar patterns of physiological activity to typically developing persons, but that individuals with ASD and a language delay, had significantly different physiological reactions to stimuli than these. The presence of co-occurring disorders may also lead to differential physiological activity among persons with autism. Recent studies have suggested that the presence of a co-occurring disorder such as fragile x syndrome (Cohen et al., 2015), or an anxiety disorder (Hollocks et al., 2014), may result in differential physiological activity than is observed among individuals with autism and no co-occurring diagnoses. Future research which examines the impact of participant characteristics such as these may provide a clearer understanding of the variables influencing physiological activity among those with ASD.

There may be difficulties in relating changes in cortisol to specific occurrences for challenging behaviour given the time taken for changes in cortisol to occur and the

obtrusive nature of the collection of salivary, plasma, or urinary cortisol for which it would be necessary to interrupt occurrences of challenging behaviour. However, although the co-activation of the LHPA axis and the ANS is not always observed (e.g., van Goozen et al., 1998), studies examining indices of ANS activation such as HR or EDA, which can be measured from a distance and with minimal interference or disruption, may yield a better understanding of the relationship between physiological activity and repetitive, stereotyped behaviour. Preliminary research in this area (e.g., Hirstein et al., 2001; Lewis et al., 1984; Lydon et al., 2013) indicates that the measurement of physiological activity surrounding stereotyped behaviour is both feasible and may contribute to our understanding of behavioural function.

Future research in this area may also benefit from the recruitment of participants with ASD based upon their engagement (high/low) in specific challenging behaviours under examination (e.g., stereotypy, SIB, aggressive/destructive behaviour). It is possible that failure to show a relationship in the current study between SIB or aggressive/destructive behaviour and cortisol may be attributed to the relatively low frequency and severity of SIB observed among participants.

Future research examining the correspondence between overt signs of stress and anxiety, such as those measured by the SSS, and physiological measures of stress would also be of use. A lack of correspondence between such measures may be considered a recommendation for more frequent physiological monitoring among individuals with ASD in order to ensure psychological and emotional wellbeing and to identify environmental or routine changes associated with distress that can be altered. The aim of the next study (Study 3: Chapter 4) was to further explore the relationship between physiological activity and stereotyped motor behaviour among persons with autism, and also to examine physiological activation during various mood states in order to assess the correspondence between overt presentation and physiological activity in this population.

Chapter 4: Study 3

Heart Rate Measurement during Stereotyped Motor Behaviour in Autism Spectrum Disorder[4]

[4] This chapter has been accepted for publication: Lydon, S., Healy, O., Mulhern, T., & Hughes, B. M. (2015c). Heart Rate Measurement during Stereotyped Motor Behavior in Autism Spectrum Disorder. *Journal of Developmental and Physical Disabilities, 27,* 677-699. doi: 10.1007/s10882-015-9445-1

Abstract

Previous research has suggested that a relationship may exist between physiological arousal and engagement in motor stereotypy among children and adolescents with autism. It has been speculated that levels of, or alterations in, physiological arousal may act as antecedents or reinforcing consequences for stereotypy. The current study sought to investigate the relationship between these two variables among five children aged between four and 17 years who were diagnosed with autism spectrum disorders. The results revealed little association between physiological arousal and stereotypy among these participants. However, a consistently atypical physiological response to stress, suggestive of physiological blunting, was observed. The implications of these findings for our understanding of the function of stereotypy, and stress responsivity among persons with autism, are discussed along with suggestions for future research.

The findings from Study 2 (Lydon et al., 2015b/Chapter 3) were suggestive of a relation between physiological activity and engagement in stereotyped motor behaviour among persons with autism. The current study aimed to further explore this relationship among a sample of children with ASD who engaged in high rates of stereotyped motor behaviour. Stereotyped, repetitive motor movements comprise one of the core diagnostic criteria for ASD (American Psychiatric Association, 2013). For this reason, it is unsurprising that such behaviours are pervasive among persons with the disorder; Campbell and colleagues (1990) examined the prevalence of stereotyped motor movements among 224 children with autism and found that almost all participants (99.5%) presented with at least mild motor stereotypy. These behaviours have also been shown to present more frequently and severely among individuals with autism than among their typically developing peers or peers with other developmental disabilities (Bodfish et al., 2000; Macdonald et al., 2007). While not physically harmful, research has demonstrated such behaviours to negatively impact on academic engagement and learning (Koegel & Covert, 1972; Morrison & Rosales-Ruiz, 1997), and social interactions and social inclusion (Cunningham & Schreibman, 2008; Durand & Carr, 1987; Loftin, Odin, & Lantz, 2008).

The cause of these behaviours is as yet unestablished (for review of recent research on this topic see Leekam, Prior, & Uljarevic, 2011). However, a relationship between physiological arousal and engagement in stereotyped behaviour has been proposed (e.g., Hutt et al., 1964; Hutt & Hutt, 1968; Lydon et al., 2013; Sugarman et al., 2014). Early theories suggested that individuals with autism experienced physiological hyper-arousal, including excessive SNS activity and significantly elevated physiological arousal, which led to many of the abnormal behaviours associated with ASD, including withdrawal, increased behavioural reactivity, and engagement in stereotyped behaviour in an attempt to block external stimulation (Hutt et al., 1964; Hutt et al., 1968). Other researchers (e.g., Rimland, 1964) alternatively suggested that individuals with ASD experience

physiological hypo-arousal and that behaviours such as stereotypy can be understood as attempts to self-stimulate and increase physiological arousal. Later, Lydon et al. (2013) suggested that stereotyped behaviours might function to allow individuals with ASD to access a preferred state of elevated physiological arousal. Most recently, Sugarman and colleagues (2014) have put forward the autonomic dysregulation theory of autism which suggests that impairments in autonomic regulation underlie these behaviours and that stereotyped behaviours may be understood as attempts to reduce stress, or high levels of physiological arousal, and to restore homeostasis for persons with autism.

Over the course of five decades, studies that have empirically examined the relationship between various indices of physiological arousal and stereotypy among individuals with autism have resulted in conflicting findings. Sroufe and colleagues (1973) examined the association between stereotypy and HR for a young boy diagnosed with ASD. Their physiological assessment suggested that stereotypy was typically preceded by a decrease in HR and followed by a significant increase in HR. The further analysis of co-occurring muscular tension and facial expression, along with the situations in which stereotypy was exhibited, suggested that stereotypy served multiple functions for the participant including the elevation of physiological arousal during periods of low environmental stimulation, the release of excess excitement or energy at other times, or the prevention of additional stimulation during periods of high physiological arousal. Hutt and colleagues (1975) investigated HR patterns during the stereotyped behaviour of nine children diagnosed with ASD. When compared to typically developing children, the participants with ASD evidenced greater SNS activity, suggestive of physiological hyper-arousal. Hutt et al.'s HR analysis suggested that the repetitive stereotyped behaviour exhibited by participants functioned to decrease physiological arousal and to block further environmental stimulation. Young and Clements (1979) examined the relationship between HR and two forms of stereotypy, hand movements and body rocking, among three youths

diagnosed with intellectual disabilities. The authors sought to determine whether stereotypy functioned to increase or decrease physiological arousal. Their results suggested that these behaviours were arousal-inducing as HR increased during their occurrence. However, HR variability was found to increase during body rocking but to decrease during occurrences of hand stereotypy suggesting that, even for the same individual, not all topographies of stereotypy may have the same physiological effects. Young and Clements suggest that diagnosis may impact upon the function, and physiological correlates, of stereotypy proposing that individuals with intellectual disabilities are more likely to engage in arousal-inducing stereotypy while individuals with ASD are more likely to exhibit de-arousing stereotypy. Willemsen-Swinkels and colleagues (1998) examined HR waveforms surrounding the stereotypy of 18 children diagnosed with various developmental disabilities including pervasive developmental disorders, language disorders, and ADHD. HR patterns that co-occurred with participants' stereotypy varied according to participants' mood state; stereotypy that was associated with happiness was preceded and followed by HR increases suggesting that such occurrences functioned to release excitement or to express joy. Stereotypy associated with distress occurred when physiological arousal was elevated and led to decreases in arousal suggesting that such occurrences functioned to soothe or calm and to block external stimulation. Stereotypy associated with outward calm was not associated with specific HR patterns and the authors posited that such occurrences might have social functions. Lydon and colleagues (2013) examined the HR patterns surrounding the stereotypy of three youths diagnosed with ASD. Their results indicated that stereotypy typically led to increases in physiological arousal for all three participants; HR increases preceding and following stereotypy were observed for two participants while an increase in HR for the third participant was observed only following stereotypy. These results tentatively suggest that such behaviours served a self-stimulatory, or arousal-increasing function, for all three participants.

Other studies have also identified associations between the activation of the LHPA axis and stereotyped behaviour. Gabriels and colleagues (2013) found that children with autism who engaged in high levels of repetitive behaviours had significantly greater physiological activation, as per measures of cortisol than children with autism who engaged in low levels of repetitive behaviours. Conversely, the data of Lydon et al. (2015b/ Study 2: Chapter 3) suggested that individuals who engaged in higher levels of stereotypy had higher cortisol levels, indicative of greater physiological arousal, than their peers who engaged in little or no stereotyped behaviour. The results of this body of research are suggestive of some association between physiological activity or activation and stereotyped behaviours. However, the conflicting results heretofore observed in the research literature highlight the need for further research to identify the nature of this relationship, possible mediators of this relationship, or personal characteristics that may be associated with this relationship.

Research into the psychophysiology of stereotypy may have important implications for the field of ABA. Incorporating measures of physiological arousal during repetitive behaviour may further our understanding of such challenging behaviour, an issue for which ABA is frequently criticised (e.g., Duerden et al., 2012; MacLean et al., 1994; Rapp & Vollmer, 2005b). For instance, Barrera and colleagues (2007) have previously noted that behaviour analysts' ability to treat challenging behaviour far outweighs our understanding of such behaviours. The examination of the role physiology may play in the development or maintenance of such behaviour may help address such criticism by furthering our knowledge about the etiology of such behaviours. Research in this area may also help with the improvement of functional assessment or analysis techniques which are considered the "gold standard" in determining behavioural function and developing treatment plans (Vollmer, Bosch, Ringdahl, & Rapp, 2014). Examining the addition of psychophysiological measurement to such assessment methods may provide a contribution

to the elucidation of behavioural function and the ability to develop precise function-based interventions for such behaviours. Romanczyk and Gillis (2006) previously noted the conceptual validity of functional analysis or assessment that incorporates physiological measurement but highlighted that further data on the clinical validity of such practices is required. It is also possible that psychophysiological assessment may further our understanding of, and ability to treat, automatically reinforced stereotyped behaviour. Automatic reinforcement describes the process by which a behaviour is maintained by the internal stimulation or sensation that it produces (Vollmer et al., 2014). Such behaviours are often referred to as self-stimulatory. Currently, the identification of a behaviour as automatically reinforcing is of limited use for treatment development (Cunningham & Schreibman, 2008; LeBlanc, Patel, & Carr, 2000; Vollmer, 1994) while the identification of the internal variables which are positively or negatively reinforcing the targeted challenging behaviour may allow for the development of function-based, effective behavioural treatments. The suggestion that the identification of a relationship between physiology and challenging behaviour may aid with treatment development is supported by the previous success of arousal-inducing, such as physical exercise (Lang et al., 2010), or de-arousing strategies, such as relaxation training (Mullins & Christian, 2001; To & Chan, 2000), in treating challenging behaviour.

The current study sought to further our understanding of the function of stereotypy among children and adolescents diagnosed with ASD. Specifically, it aimed to develop a practitioner-friendly model for the assessment of the association between specific HR patterns and engagement in stereotypy, and for the analysis of the resulting data. This model was implemented in the assessment of the stereotypy of five youths diagnosed with ASD. Further, this study sought to expand on previous research in the area by examining whether any co-occurring physiological waveforms differed according to participant mood

state during engagement in the behaviour. Finally, it sought to examine the level of HR according to participant mood state.

Method

Ethical Approval, Participants, Target Behaviours, and Setting

The study was approved by an ethical research review board at the National University of Ireland Galway. Consent was sought first from parents or caregivers, and then participants where possible.

Five individuals diagnosed with ASD who exhibited high rates of stereotyped motor behaviours were recruited to participate in this study. None of the participants were diagnosed with a medical condition, or taking medication at the time of the study, that may have affected their HR responses.

Participant 1 was a 5-year-old male diagnosed with autism. His weight was 17.6 kg and his height was 102 cm. He had been diagnosed with autism at four years of age, and had begun receiving early intervention, based on the principles of ABA, at the age of 3 year 10 months. Participant 1 was verbal but engaged in little spontaneous communication with others. A recent psychological assessment, which included administration of the Wechsler Pre-School and Primary Scale of Intelligence and the Adaptive Behavior Assessment System (3rd Ed.), indicated that Participant 1 scored within the low average range of intelligence while his adaptive behaviour and independent skills were significantly impaired and not within an age-appropriate range. Both Participant 1's parents and teachers reported that he engaged in stereotyped motor behaviour near-constantly and that these behaviours interfered greatly with his ability to interact socially and with his academic progress. The topographies of Participant 1's stereotypy are presented in Table 9. The results of a Questions about Behavioral Function (QABF; Matson & Vollmer, 1995), an indirect functional assessment instrument, completed by the Board Certified Behaviour Analyst® at Participant 1's school, indicated that these

behaviours were multiply controlled with both non-social (i.e., maintained by sources of automatic reinforcement) and escape functions. Participant 1's QABF data, along with those of the other participants in this study, are presented graphically in Appendix A. Discrimination training had been introduced in an attempt to reduce his stereotyped behaviour but had failed to produce significant decreases in these behaviours. This intervention was discontinued during the data collection period of the current study.

Table 9

Topographies and Operational Definitions for the Stereotypy of all Participants

Topography	Operational Definition	P1	P2	P3	P4	P5
Head Stereotypy						
Chin pushing	raising chin upwards with fingers or palm					✓
Ear pressing or covering	pressing one finger into one or both ears, or pressing fist or open palms against both ears simultaneously			✓		
Face covering	placing hands flat against eyes or face					✓
Facial grimacing	scrunching or contorting of the face or nose	✓	✓			
Hair rubbing	grasping, rubbing, or tugging on hair		✓	✓		
Head nodding	moving head up and down quickly without communicative intent			✓		
Head shaking	turning head from side to side quickly without communicative intent		✓	✓		
Repetitive teeth movements	clenching or rubbing of teeth together or engagement in functionless, repetitive biting movements with teeth	✓	✓			
Teeth touching	pressing one or more fingers against teeth					✓
Tongue movements	curling tongue and pushing it through pursed lips or pushing tongue out of mouth and letting it hang over lips	✓	✓		✓	
Hand and Finger Stereotypy						
Finger sucking	placing finger in mouth and closing lips around it				✓	
Hand clapping	hitting hands eith together, while flat or in fists, or on a surface	✓	✓			✓
Hand flapping	moving one or both hands up and down quickly	✓	✓	✓	✓	✓
Hand squeezing	Grasping or squeezing one hand within the other	✓	✓		✓	✓
Hand pressing	Pushing palms together forcibly	✓				✓
Hand rubbing	brushing hands together or rubbing skin, body parts, or objects with open palm(s)	✓		✓	✓	✓
Lip pressing	pushing finger against lips				✓	
Repetitive finger movements	clenching fingers together, wiggling fingers, tapping nose or other objects with fingers	✓	✓	✓	✓	
Skin and nail picking	pulling or digging at skin on fingers, pulling under fingernails with other fingernail or object, biting and pulling skin on fingers			✓		
Skin pulling	clenching skin between fingers and tugging it out of place. Can be skin anywhere on body including near eyes					✓
Body Stereotypy						
Body bouncing	moving up and down quickly while standing, seated, or moving	✓		✓	✓	✓
Body rocking	tilting body backwards and forwards in fast or slow motion	✓	✓	✓	✓	✓
Body writhing	contorting body into atypical positions or writhing body				✓	✓
Jumping	leaping of the ground when walking or running	✓			✓	
Limb swinging	moving limbs (either arms or legs) back and forth quickly		✓	✓	✓	✓
Pacing	walking or running back and forth in the same steps	✓	✓			
Repetitive foot movements	swinging feet while sitting, stamping feed on the ground	✓	✓		✓	✓
Shoulder shrugging	moving shoulders up and down quickly			✓		
Stereotyped object manipulation	squeezing or repetitively tapping objects	✓	✓	✓	✓	

Participant 2 was a 4-year-old male diagnosed with ASD. His weight was 17.7 kg and his height was 99 cm. Participant 2 had been diagnosed at the age of 2 years and 6 months and had begun receiving an ABA educational intervention at the age of 3 years and 10 months. Recent assessments, including the Vineland Adaptive Behavior Scales (2nd Ed.) and the Griffiths Mental Development Scales, indicated that Participant 2 had a low level of functioning across communication, social, motor, and independence domains. Participant 2 exhibited multiple topographies of stereotypy (see Table 9) that occurred at high rates and were reported to interfere with instructional sessions. A QABF completed by the school's Board Certified Behaviour Analyst® indicated that these behaviours were primarily non-social but that some instances appeared to serve an escape function. No behavioural interventions had been implemented for his stereotypy.

Participant 3 was a 17-year-old male diagnosed with autism and a mild intellectual disability. His weight was 56.7 kg and his height was 160 cm. Participant 3 had been diagnosed with autism at the age of 3 years. He had received early intensive behavioural intervention as a child and was currently attending a special school for children and adolescents with ASD. Participant 3 was verbal but rarely engaged in spontaneous communication with others. He engaged in challenging behaviour in the form of near-constant vocal and motor stereotypy that was reported to impede his academic and social engagement. The topographies of motor stereotypy in which Participant 3 engaged are presented in Table 9. A QABF completed by Participant 3's teacher suggested that these behaviours were multiply controlled, serving primarily a non-social function but also functioning to provide a means of escape, as an indicator of physical discomfort, and to access social attention. No behavioural interventions had been implemented for his stereotypy.

Participant 4 was a 9-year-old male diagnosed with autism and a mild intellectual disability. His weight was 27.9 kg and his height was 137.2 cm. Participant 4 had advanced

communication and social abilities. However, he engaged in high rates of motor stereotypy that were reported to hinder his social interactions with peers and to impact upon his engagement with academic tasks. The topographies of Participant 4's motor stereotypy are presented in Table 9. A QABF completed by Participant 4's teacher indicated that these behaviours were primarily non-social and also functioned as a means to access social attention although to a lesser extent. No behavioural interventions had been implemented for this participant's stereotypy.

Participant 5 was a 10-year-old male diagnosed with autism and a mild intellectual disability. His weight was 34.5 kg and his height was 139.7 cm. Participant 5 was verbal but rarely engaged in spontaneous verbal exchanges with others. Participant 5 engaged in multiple forms of motor stereotypy (see Table 9) but no intervention had been implemented for these previously. The results of a QABF completed by Participant 5's teacher indicated that his motor stereotypy was primarily automatically reinforced but also served, on occasion, as an indicator of physical pain or discomfort.

Data collection for Participants 1 and 2 occurred at their school, an early intervention service that delivered ABA educational interventions, while data collection for Participants 3, 4, and 5 occurred at a special school for children and adolescents that adhered to an eclectic model of education and intervention for autism.

Behavioural Recording

Data were collected on each occurrence of the target behaviours. These data included the time of onset and offset of the behaviour, the ongoing activity, behavioural intensity, and mood state. Behavioural intensity was rated as either: *mild*, defined as behaviours that occurred in the absence of vocalisations and involved slow and discrete motor movement; *moderate*, defined as behaviours that were accompanied by vocalisations which were lower than or of normal speaking volume and/or the accompanying motor movements were loose, not rigid, and the speed and degree of movement approximated

that of appropriate motor movements; or *severe*, defined as behaviours that were accompanied by vocalisations which were notably louder than usual speaking volume and/or the accompanying motor movements were vigorous and quick, and/or markedly rigid. Mood state was recorded utilising the guidelines outlined by Willemsen-Swinkels and colleagues (1998). Observers recorded whether target behaviours occurred during "elation", "distress", or "composure" on the basis of overt signs of emotion including vocalisations, verbal expressions, facial expressions, or behaviours other than the target stereotyped behaviour patterns (e.g., tantrum behaviours; see Willemsen-Swinkels et al., 1998).

An interobserver was present during two of the five days of data collection for each participant. Interobserver agreement was assessed across the five categories of behavioural recording (i.e., onset time, offset time, ongoing activity, behavioural severity, and mood state). The data recorded for the time of onset and time of offset for each behaviour were considered to agree if the recording differed by no more than two seconds. Agreements for the remaining categories were defined as an exact match in the behavioural recording of the observers. Interobserver agreement (presented in Table 10) was calculated by dividing the total number of agreements by the total number of opportunities for agreement.

Table 10

Interobserver Agreement for Behavioural Recording conducted for Participants in Study 3

Participant	Mean IOA for behavioural recording	Onset time	Offset time	Ongoing activity	Behavioural severity	Mood state
1	97.2%	95.1%	93.7%	100%	100%	97.2%
2	96.5%	96.4%	93.8%	99%	96%	97.5%
3	93.8%	92.2%	90.2%	95.8%	95.6%	95.3%
4	95.8%	95.4%	92.9%	97.1%	96.7%	96.7%
5	97.6%	97.9%	96.3%	100%	97.5%	96.3%

Note: IOA= interobserver agreement.

Heart Rate Recording

HR recording was conducted using the Polar RS800cx (Polar Electro OY, Kempele, Finland). This device consists of a chest strap and wristwatch. The chest strap is worn around the ribcage where it records the electrical signal of the heart every 1 second. HR data are then transmitted to, and stored by, the wristwatch which was worn either on participants' wrists (Participants 1, 3, 4, and 5) or attached to the belt loop of their trousers in order to reduce distraction (Participant 2). Physiological monitoring in the current study was conducted in accordance with best practice for the inclusion of children with autism in psychophysiological experiments (see Kylliainen, Jones, Gomot, Warreyn, & Falck-Ytter, 2014). While these guidelines relate primarily to laboratory-based experiments involving the presentation of experimental stimuli, relevant recommendations including the use of a preliminary desensitisation (habituation) period, consultation with parents and teachers about participants' likely responses to the HR monitor and ways of facilitating their comfort with it, the use of behavioural strategies including positive reinforcement to encourage wearing of the HR monitor, and the reliance on clinically experienced researchers to conduct data collection were incorporated into our methodology.

While the use of a single measure of physiological activity has been criticised (Romanczyk & Matthews, 1998), the decision to use a HR measure alone was made on the basis of previous research in the area which has evidenced positive findings with the use of HR measures (e.g., Freeman et al., 1999; Lydon et al., 2013; Willemsen-Swinkels et al., 1998) and consideration of the practical difficulties of applying more than one physiological recording device to participants diagnosed with ASD. Polar HR monitors have been previously used with individuals diagnosed with ASD (e.g., Freeman et al., 1999; Hodgetts, Magill-Evans, & Misiaszek, 2011; Lydon et al., 2013; Willemsen-Swinkels et al., 1998) and their suitability for recording HR in applied settings has also been emphasised (Laukkanen & Virtanen, 1998). Previous research has also demonstrated

the reliability of Polar HR monitors, when compared to ECG, to be adequate (Laukkanen & Virtanen, 1998; Terbizan, Dolezal, & Albano, 2002).

Experimental Design and Procedure

The current study was conducted using a naturalistic, single-subject research design. Thus, all data were collected in participants' typical service setting and with minimal interference to their usual routines or schedules. Participants were habituated to wearing the HR monitor for two days prior to data collection. Habituation involved showing participants the chest strap and wristwatch, describing its purpose in simple terms, allowing participants to ask questions, and where necessary gradually applying the HR monitor to the participants' chest area. Participants received access to reinforcing items or activities contingent on their wearing the HR monitor during the habituation period. The operational definitions for the topographies of stereotypy exhibited by each participant, presented in Table 9, were also developed by the researcher during each participant's habituation phase and subsequently used by the observer(s) to record the occurrence of stereotypy during data collection. Data collection, including behavioural and HR recording, subsequently occurred across a period of five school days. During this time, participants were fitted with the HR monitor each morning and the observer(s) recorded the occurrence of each target behaviour across the school day. Observer(s) used a stopwatch, synced each morning with the Polar RS800cx wristwatch to ensure correspondence between the HR and behavioural record, to record the onset and offset of each target behaviour. As target behaviours were near-constant in some cases, a 5 second period during which all target behaviours were absent was considered to constitute an end to an occurrence of a target behaviour.

Data Analysis

An extensive review of the literature assessing the relationship between physiological activity and engagement in challenging behaviour was conducted prior to

this study, with a focus on the data analysis methodologies employed by previous research studies in this area. Furthermore, the data analysis methodologies recommended for use in psychophysiologal studies of challenging behaviour by Cohen et al. (2011) were considered. Such methodologies included sequential analysis procedures, time series analysis, multi-level modelling, and survival analysis (see Cohen et al. for a description of appropriate modes of employment). These methodologies were carefully considered and discussed among the research team and also with a number of practicing behaviour analysts. A concensus was reached that there was a problematic disconnect between research and practice in this area; while research demonstrated established means of assessing the relationship between physiological activity and engagement in challenging behaviour, these did not appear to be utilised in many applied settings. This was considered problematic given the emergence, and popularity, of non-function based interventions for challenging behaviour that were based upon an assumed, but not pre-determined, relationship between physiological arousal and challenging behaviour (see: McDonnell et al., 2015). Thus, a key aim of the current research study was to develop a model of assessment and data analysis that could easily be applied by a practicing behaviour analyst. It is known that the use of complex statistical techniques can alienate clincians and reduce their engagement with, and understanding of research (e.g., Cohen, Sargent, & Sechrest, 1986; Herbert, 2003; Stewart & Chambless, 2010; Stewart, Shirman, & Chambless, 2012). Thus, the data analysis methodology selected, and outlined below, was considered to be an appropriate and effective way of assessing the association, if any, between physiological activity and engagement in challenging behaviour. It also yielded a mode of analysis that did not require practitioners to have a high degree of statistical knowledge or competency or access to specialised statistical programs.

As participants' stereotyped behaviour occurred at such high frequencies during the data collection period, it was not possible to analyse every single occurrence. For this

reason, the data were sampled as follows: every fourth incident of stereotypy during each mood state (i.e., elation, distress, and composure) was analysed. Exceptions to this were if stereotypy relating to any mood state occurred less than 60 times or if occurrences were rated as being of severe behavioural intensity. In such cases, the HR data for all incidents of stereotypy were analysed. If data for a sampled incident was missing, then the next incident occurring during the same mood state was analysed in its place. Finally, incidents that included disagreements in the onset time, offset time, or mood state recorded by observers were excluded from the analysis of HR data.

The association between stereotypy and changes in HR was first assessed for occurrences of stereotypy relating to each mood state individually and for all occurrences of stereotypy following this. The first step of the analysis for each occurrence of stereotypy was to assess for the presence of HR changes surrounding the behaviour's onset in order to determine whether changes in HR may have served as an antecedent for the behaviour. To do this, mean HR during the 10 seconds preceding the onset of the behaviour was compared to HR during the 10 seconds following the onset of the behaviour, or the maximum period possible in cases of shorter occurrences of stereotypy. Any change equivalent to or greater than the SD of the 10 seconds preceding the onset of the behaviour was considered to be a significant change (i.e., the episodic measure of HR was considered to deviate significantly from that observed during baseline). The SD of baseline HR was used as a threshold for inferring significant changes as it encapsulates the anticipated spread of typical measures of HR during rest (i.e., the range within which HR could be expected to vary should it *not* change). Thus, subsequent measures of HR that fell outside the range of baseline ±1 SD were inferred to have deviated significantly away from baseline. For each occurrence of stereotypy, the HR patterns surrounding behavioural onset observed were subsequently categorised as no change (i.e., change in HR is less than or equivalent to the SD of HR during the baseline period), significant increase (i.e., increase

in HR that is greater than the value of the SD of HR during the baseline period), or significant decrease (i.e., decrease in HR that is greater than the value of the SD of HR during the baseline period). This analysis was repeated in order to compare HR during the 10 seconds prior to the offset of stereotypy to HR during the 10 seconds following the offset of the behaviour in order to identify any HR patterns occurring around the offset of the behaviour that may have functioned as reinforcement for the behaviour (i.e., increases or decreases in physiological arousal). For this analysis, HR change was interpreted using the SD of the HR measures during the 10 seconds prior to the offset, or the maximum period possible for shorter occurrences of stereotypy. Once again, the HR pattern observed was subsequently categorised as no change, increase, or decrease. Finally, this analysis was performed to assess the shift in HR as a result of the behaviour. This involved the comparison of HR level during the 10 seconds prior to the onset of the behaviour and HR level during the 10 seconds following the offset of the behaviour. HR change was interpreted with regard to the SD of the HR measures during the 10 second period prior to the occurrence of the behaviour.

Analyses were conducted for each sampled occurrence allowing for the categorisation of HR patterns at onset and offset, and the overall shift in HR level as a result of the occurrence of stereotypy. These analyses were also performed for an equal number of non-occurrences. Non-occurrences were defined as a 30 second period during which no stereotyped behaviour was emitted that were preceded and followed by 30 seconds during which no challenging behaviour occurred. These incidents were used as a comparison in order to examine whether the frequency with which the various possible HR patterns occurred during incidents of stereotypy deviated from what was observed during non-occurrences of the behaviour. Using data from occurrences and non-occurrences, a frequency table was prepared which listed that number of incidents categorised as no change, increase, or decrease for each of the three assessments (i.e., patterns at onset,

patterns at offset, overall shift in HR level) for both occurrences and non-occurrences of stereotyped behaviour. Following this, a chi square analysis was performed for each assessment point to determine whether the frequency of occurrence of each HR pattern was evenly spread for both occurrences and non-occurrences. For this analysis, we tested against the null hypothesis that no changes, increases, and decreases would occur at similar frequencies.

Following this, mean HR throughout incidents of stereotypy occurring during composure, elation, and distress mood states was assessed. To achieve this, mean HR was calculated for each 12 second epoch within each occurrence of stereotypy. The mean of all 12 second epochs falling within occurrences of stereotypy during each mood state was subsequently calculated, yielding an overall mean HR for each mood state. Mean HR during each mood state was subsequently graphed for each of the five days of measurement for each participant. Visual inspection was used to determine whether consistency in the level of HR during each mood state across the days of measurement was discernible for each participant. In addition to visual inspection, and to illustrate the nature of the effects, the data for each participant were scrutinised using Analyses of Variance treating each data-point as a separate case. For each participant, these data comprised several hundred individual measures for a sample of n = 1; treating each data-point as a separate case was akin to conducing an ANOVA on a sample of several hundred participants yielding one measure each. This allowed the computation of indicative p-values relating to differences in observations from the different mood states, with which to bolster conclusions drawn from visual inspections. As ANOVA is based on an assumption of independent measures across multiple participants, the resulting p-values should be viewed as analogous to actual statistical significance levels; albeit in a way that benefits from the repeated-measures nature of the dataset. One-way between subjects ANOVAs

with HR as the dependent variable were conducted. Mood state with three levels (i.e., composure, elation, distress) served as the independent variable in these analyses.

Results

Characteristics of Motor Stereotypy

Behavioural observation and HR recording took place across 18 hours and 56 minutes for Participant 1. Across the five days of recording, 385 incidents of stereotypy were observed. The total duration of all occurrences observed was 13.2 hours (*M*= 123.47 seconds, range= 1 second-16.95 minutes), 69.8% of the total recording time. Of these occurrences, 42.9% were rated as being of mild behavioural intensity, 52.7% were rated as being of moderate behavioural intensity, and 4.4% were rated as being of severe behavioural intensity. With regards mood state, stereotypy occurred most frequently during composure (58.4%; 59.2% of incidents rated as mild, 36.4% rated as moderate, 4.4% rated as severe), followed by elation (30.1%; 24.7% of incidents rated as mild, 74.3% rated as moderate, 0.9% rated as severe), and distress (11.4%; 86.4% of incidents rated as moderate, 13.6% rated as severe). Stereotypy occurred with an equal frequency during leisure time and instructional time (37.9%), and was less frequent during mealtimes (9.1%), transitioning (7.8%), group activities (0.8%), and other activities (6.5%).

Data collection for Participant 2 occurred across 16 hours 51 minutes and 26 seconds. There were 400 occurrences of stereotypy during the recording period. The total duration of stereotyped behaviour that occurred during this period was 5.4 hours (*M*= 46.93 seconds, range= 1 second-11.85 minutes) or 32% of the total recording time. Occurrences were most frequently rated as being of moderate behavioural intensity (52%), followed by mild behavioural intensity (37.8%), and severe behavioural intensity (3.5%). Stereotypy occurred most frequently while the participant appeared overtly elated (60%; 38.6% of occurrences rated as mild, 60.2% rated as moderate, and 1.2% rated as severe), while stereotypy during periods of composure (37.8%; 61.4% of occurrences rated as mild,

34.5% of occurrences rated as moderate, 4.1% of incidents rated as severe) was less frequent, and stereotypy during periods of distress occurred rarely (2.3%; 77.8% of incidents rated as moderate, 22.2% of incidents rated as severe). Stereotypy was most prevalent during leisure time (57.8%), followed by instructional time (15.3%), mealtimes (15%), transitioning (6.5%), group activities (4%), and 12.5% of occurrences were during other activities.

Data collection for Participant 3 occurred across 18 hours 22 minutes and 35 seconds. Across the five days of recording, 596 occurrences of motor stereotypy were recorded. The total duration of stereotyped behaviour was 7 hours 59 minutes and 17 seconds, or 43.5% of the total recording time. The mean duration of an occurrence of stereotypy was found to be 48 seconds (range 0 seconds- 35.04 minutes). Occurrences were most frequently rated as mild (99%), and less frequently as moderate (1%). Stereotypy most commonly occurred during composure (87.8%; 100% of incidents were mild), followed by elation (9.8%; 100% of incidents were mild), and then distress (2.5%; 53.5% of incidents were rated as mild, 46.7% were rated as moderate). Stereotypy was most prevalent during group work or group activities (66.4%), followed by leisure activities or free time (16.3%), eating (6.2%), 1:1 work (6%), and other activities (5%).

Data collection for Participant 4 occurred across 20 hours 10 minutes and 52 seconds. Over the recording period, 436 incidents of motor stereotypy were recorded. The total duration of stereotypy observed was 8 hours 45 minutes and 11 seconds, or 43.4% of the total recording time. The mean duration of an occurrence was 75 seconds (range 1 second- 19.38 minutes). Observer(s) most frequently rated stereotypy as being mild (77.1%), and less frequently as moderate (22.9%). The majority of occurrences took place during composure (68.3%; 91% of incidents were rated as mild, 9% of incidents were rated as moderate), followed by elation (24.1%; 65.8% of incidents were mild, 34.2% of incidents were moderate), and distress (7.6%; 27.3% of incidents were rated as mild,

72.7% of incidents were rated as moderate). Stereotypy was most prevalent during group work or group activities (40.4%), followed by independent work (27.3%), free time or leisure activities (15.1%), eating (12.4%), transitioning (4.6%), and other activities (0.5%).

Data collection for Participant 5 occurred across 19 hours 31 minutes and 4 seconds. In total, 391 occurrences of motor stereotypy were recorded. The total duration of occurrences of stereotypy observed was 6 hours 36 minutes and 44 seconds, or 33.9% of the total recording time. The mean duration of occurrences of stereotypy was 61 seconds (range 2 seconds-11.25 minutes). Occurrences of stereotypy were most frequently rated as being mild (68.5%), moderate (31.2%), or severe (0.3%). Participant 5 most frequently engaged in stereotypy during periods of composure (60.5%; 89.2% of incidents rated as mild, 10.8% of incidents rated as moderate) or elation (34.5%; 26% of incidents rated as mild, 73.2% of incidents rated as moderate, 0.8% of incidents rated as severe). No occurrences of stereotypy during distress were observed during the recording period. Engagement in stereotypy was most frequent during group work or group activities (49.6%), followed by free time or leisure activities (20.2%), independent work (10.5%), eating (8.7%), transitioning (7.2%), 1:1 work (3.6%), and other activities (0.3%)

Cardiovascular Data Analysis

Participant 1's mean HR was found to be 121.39 bpm (*SD*= 10.87; range 78-193 bpm). HR patterns surrounding occurrences of stereotypy were subsequently examined. The outcomes of this analysis for occurrences of stereotypy during composure are presented in the upper left panel of Table 11. The upper left panels of Table 12 and Table 13 display the data for occurrences of stereotypy during elation and distress respectively. It was not possible to identify any HR pattern, at either onset or offset, or a shift in HR associated with engagement in stereotypy, that was significantly more likely to be associated with occurrences of stereotypy than with non-occurrences of stereotypy during any of the three mood states. HR patterns across all occurrences of stereotypy were

subsequently analysed. The results of this analysis for Participant 1 are presented in the upper left panel of Table 14. An increase in HR at the onset of stereotypy was significantly more likely to occur than at the onset of non-occurrences. There were no HR patterns observed to occur consistently at the offset of stereotypy nor was the overall shift in HR consistent for this participant. Following this analysis, the mean level of HR during occurrences of stereotypy associated with each of the three mood states was examined. The results of this analysis are presented in the upper left panel of Figure 3. Visual analysis revealed that HR was highest during occurrences of stereotypy associated with elation, lower during composure, and lowest during occurrences associated with distress. This finding was subsequently explored further using a one-way between subjects ANOVA. This analysis revealed that the differences in mean HR between stereotypy occurring during different mood states, $F(2,1899)=84$, $p=.001$, would be statistically significant under appropriate conditions. A Tukey post-hoc test revealed that HR was significantly higher during elation ($M=126.31$, $SD=11.95$) than during composure ($M=120.72$, $SD=9.15$), $p<.001$, higher during elation than during distress ($M=119.37$, $SD=7.79$), $p<.001$, and higher during composure than distress, $p<.05$.

Table 11

Heart Rate Patterns associated with Occurrences of Stereotypy during Composure for Participant 1 (upper left panel), Participant 2 (upper middle panel), Participant 3 (upper right panel), Participant 4 (lower left panel), and Participant 5 (lower right panel)

	Participant 1			**Participant 2**			**Participant 3**	
	Occurrences of Stereotypy (*n*=61)	**Non-occurrences of Stereotypy (*n*=61)**		**Occurrences of Stereotypy (*n*=38)**	**Non-occurrences of Stereotypy (*n*=38)**		**Occurrences of Stereotypy (*n*=130)**	**Non-occurrences of Stereotypy (*n*=130)**
Onset			**Onset**			**Onset**		
No change	*n*=23	*n*=34	No change	*n*=21	*n*=23	No change	*n*=61	*n*=65
Increase	*n*=21	*n*=14	Increase	*n*=10	*n*=7	Increase	*n*=38	*n*=32
Decrease	*n*=17	*n*=13	Decrease	*n*=7	*n*=8	Decrease	*n*=31	*n*=33
	$X^2(2)=4.06$, $p=.13$			**$X^2(2)=0.69$, $p=.71$**			**$X^2(2)=0.70$, $p=.70$**	
Offset			**Offset**			**Offset**		
No change	*n*=25	*n*=21	No change	*n*=14	*n*=20	No change	*n*=40	*n*=67
Increase	*n*=13	*n*=20	Increase	*n*=10	*n*=8	Increase	*n*=44	*n*=38
Decrease	*n*=23	*n*=20	Decrease	*n*=14	*n*=10	Decrease	*n*=46	*n*=25
	$X^2(2)=2.04$, $p=.36$			**$X^2(2)=2.04$, $p=.36$**			**$X^2(2)=13.46$, $p<.001$ (more likely to be no change at offset of non-occurrences)**	
Overall change in physiological arousal associated with stereotypy			**Overall change in physiological arousal associated with stereotypy**			**Overall change in physiological arousal associated with stereotypy**		
No change	*n*=21	*n*=30	No change	*n*=18	*n*=17	No change	*n*=50	*n*=51
Increase	*n*=23	*n*=17	Increase	*n*=6	*n*=8	Increase	*n*=44	*n*=45
Decrease	*n*=17	*n*=14	Decrease	*n*=14	*n*=13	Decrease	*n*=36	*n*=34
	$X^2(2)=2.78$, $p=.25$			**$X^2(2)=2.78$, $p=.25$**			**$X^2(2)=0.34$, $p=.84$**	

	Participant 4			**Participant 5**	
	Occurrences of Stereotypy (*n*=75)	**Non-occurrences of Stereotypy (*n*=75)**		**Occurrences of Stereotypy (*n*=65)**	**Non-occurrences of Stereotypy (*n*=65)**
Onset			**Onset**		
No change	*n*=32	*n*=42	No change	*n*=25	*n*=40
Increase	*n*=21	*n*=17	Increase	*n*=20	*n*=14
Decrease	*n*=22	*n*=16	Decrease	*n*=20	*n*=11
	$X^2(2)=2.72$, $p=.26$			**$X^2(2)=7.1$, $p=.03$ (more likely to be no change before non-occurrences)**	
Offset			**Offset**		
No change	*n*=30	*n*=33	No change	n=29	*n*=35
Increase	*n*=23	*n*=22	Increase	n=19	*n*=11
Decrease	*n*=22	*n*=20	Decrease	n=17	*n*=19
	$X^2(2)=0.26$, $p=.88$			**$X^2(2)=2.81$, $p=.25$**	
Overall change in physiological arousal associated with stereotypy			**Overall change in physiological arousal associated with stereotypy**		
No change	*n*=32	*n*=35	No change	*n*=30	*n*=27
Increase	*n*=25	*n*=24	Increase	*n*=15	*n*=19
Decrease	*n*=18	*n*=20	Decrease	*n*=20	*n*=19
	$X^2(2)=0.27$, $p=.87$			**$X^2(2)=0.65$, $p=.72$**	

Note. *n* refers to the number of occurrences of stereotypy or number of non-occurrences of stereotypy.

Table 12

Heart Rate Patterns associated with Occurrences of Stereotypy during Elation for Participant 1 (upper left panel), Participant 2 (upper middle panel), Participant 3 (upper right panel), Participant 4 (lower left panel), and Participant 5 (lower right panel)

	Participant 1			**Participant 2**			**Participant 3**	
	Occurrences of Stereotypy (*n*=36)	**Non-occurrences of Stereotypy (*n*=36)**		**Occurrences of Stereotypy (*n*=66)**	**Non-occurrences of Stereotypy (*n*=66)**		**Occurrences of Stereotypy (*n*=41)**	**Non-occurrences of Stereotypy (*n*=41)**
Onset			**Onset**			**Onset**		
No change	*n*=12	*n*=11	No change	*n*=19	*n*=32	No change	*n*=13	*n*=20
Increase	*n*=18	*n*=10	Increase	*n*=32	*n*=20	Increase	*n*=22	*n*=11
Decrease	*n*=6	*n*=15	Decrease	*n*=15	*n*=14	Decrease	*n*=6	*n*=10
	$X^2(2)=6.19$, $p=.05$			**$X^2(2)=1.48$, $p=.48$**			**$X^2(2)=0.70$, $p=.70$**	
Offset			**Offset**			**Offset**		
No change	*n*=15	*n*=18	No change	*n*=31	*n*=41	No change	*n*=9	*n*=21
Increase	*n*=6	*n*=11	Increase	*n*=9	*n*=13	Increase	*n*=7	*n*=10
Decrease	*n*=15	*n*=7	Decrease	*n*=26	*n*=12	Decrease	*n*=25	*n*=10
	$X^2(2)=4.65$, $p=.10$			**$X^2(2)=2.04$, $p=.36$**			**$X^2(2)=16.21$, $p<.001$ (more likely to be no change at offset of non-occurrences)**	
Overall change in physiological arousal associated with stereotypy			**Overall change in physiological arousal associated with stereotypy**			**Overall change in physiological arousal associated with stereotypy**		
No change	*n*=11	*n*=13	No change	*n*=25	*n*=29	No change	*n*=15	*n*=13
Increase	*n*=15	*n*=10	Increase	*n*=18	*n*=17	Increase	*n*=8	*n*=17
Decrease	*n*=10	*n*=13	Decrease	*n*=23	*n*=20	Decrease	*n*=18	*n*=11
	$X^2(2)=1.56$, $p=.46$			**$X^2(2)=0.53$, $p=.77$**			**$X^2(2)=0.34$, $p=.84$**	

	Participant 4			**Participant 5**	
	Occurrences of Stereotypy (*n*=35)	**Non-occurrences of Stereotypy (*n*=35)**		**Occurrences of Stereotypy (*n*=38)**	**Non-occurrences of Stereotypy (*n*=38)**
Onset			**Onset**		
No change	*n*=15	*n*=16	No change	*n*=10	*n*=22
Increase	*n*=5	*n*=12	Increase	*n*=20	*n*=11
Decrease	*n*=15	*n*=7	Decrease	*n*=8	*n*=5
	$X^2(2)=2.72$, $p=.26$			**$X^2(2)=5.8$, $p=.05$**	
Offset			**Offset**		
No change	*n*=19	*n*=19	No change	*n*=17	*n*=24
Increase	*n*=7	*n*=7	Increase	*n*=7	*n*=9
Decrease	*n*=9	*n*=9	Decrease	*n*=14	*n*=5
	$X^2(2)=0.26$, $p=.88$			**$X^2(2)=5.71$, $p=.06$**	
Overall change in physiological arousal associated with stereotypy			**Overall change in physiological arousal associated with stereotypy**		
No change	*n*=4	*n*=13	No change	*n*=10	*n*=15
Increase	*n*=8	*n*=13	Increase	*n*=20	*n*=14
Decrease	*n*=23	*n*=9	Decrease	*n*=8	*n*=9
	$X^2(2)=0.08$, $p=.96$			**$X^2(2)=12.08$, $p=.002$ (more likely to be an overall increase for occurrences)**	

Note. n refers to the number of occurrences of stereotypy or number of non-occurrences of stereotypy.

Table 13

Heart Rate Patterns associated with Occurrences of Stereotypy during Distress for Participant 1 (upper left panel), Participant 2 (upper right panel), Participant 3 (lower left panel), and Participant 4 (lower right panel)

	Participant 1 Occurrences of stereotypy (*n*=42)	Non-occurrences of stereotypy (*n*=42)
Onset		
No Change	*n*=14	*n*=18
Increase	*n*=21	*n*=15
Decrease	*n*=7	*n*=9
	$X^2(2)=1.75$, $p=.42$	
Offset		
No Change	*n*=14	*n*=22
Increase	*n*=11	*n*=7
Decrease	*n*=17	*n*=13
	$X^2(2)=3.2$, $p=.20$	
Overall change in physiological arousal associated with stereotypy		
No Change	*n*=11	*n*=11
Increase	*n*=14	*n*=14
Decrease	*n*=17	*n*=17
	$X^2(2)=0$, $p=1$	

	Participant 2 Occurrences of stereotypy (*n*=9)	Non-occurrences of stereotypy (*n*=9)
Onset		
No Change	*n*=6	*n*=4
Increase	*n*=3	*n*=3
Decrease	*n*=0	*n*=2
	$X^2(2)=2.4$, $p=.53$	
Offset		
No Change	*n*=2	*n*=4
Increase	*n*=3	*n*=1
Decrease	*n*=4	*n*=4
	$X^2(2)=1.67$, p=.44	
Overall change in physiological arousal associated with stereotypy		
No Change	*n*=4	*n*=4
Increase	*n*=3	*n*=2
Decrease	*n*=2	*n*=3
	$X^2(2)=0.40$, $p=.82$	

	Participant 3 Occurrences of stereotypy (*n*=15)	Non-occurrences of stereotypy (*n*=15)
Onset		
No Change	*n*=8	*n*=7
Increase	*n*=6	*n*=6
Decrease	*n*=1	*n*=2
	$X^2(2)=0.40$, $p=.81$	
Offset		
No Change	*n*=7	*n*=6
Increase	*n*=4	*n*=2
Decrease	*n*=4	*n*=7
	$X^2(2)=1.56$, $p=.46$	
Overall change in physiological arousal associated with stereotypy		
No Change	*n*=7	*n*=2
Increase	*n*=3	*n*=7
Decrease	*n*=5	*n*=6
	$X^2(2)=4.47$, $p=.11$	

	Participant 4 Occurrences of stereotypy (*n*=33)	Non-occurrences of stereotypy (*n*=33)
Onset		
No Change	*n*=13	*n*=10
Increase	*n*=6	*n*=11
Decrease	*n*=14	*n*=12
	$X^2(2)=2.02$, $p=.37$	
Offset		
No Change	*n*=19	*n*=19
Increase	*n*=7	*n*=9
Decrease	*n*=7	*n*=5
	$X^2(2)=0.58$, $p=.81$	
Overall change in physiological arousal associated with stereotypy		
No Change	*n*=9	*n*=13
Increase	*n*=10	*n*=11
Decrease	*n*=14	*n*=9
	$X^2(2)=1.86$, $p=.39$	

Note. n refers to the number of occurrences of stereotypy or number of non-occurrences of stereotypy.

Table 14

Heart Rate Patterns associated with Occurrences of Stereotypy for Participant 1 (upper left panel), Participant 2 (upper middle panel), Participant 3 (upper right panel), Participant 4 (lower left panel), and Participant 5 (lower right panel)

	Participant 1			**Participant 2**			**Participant 3**	
	Occurrences of Stereotypy (*n*=139)	**Non-occurrences of Stereotypy (*n*=139)**		**Occurrences of Stereotypy (*n*=113)**	**Non-occurrences of Stereotypy (*n*=113)**		**Occurrences of Stereotypy (*n*=186)**	**Non-occurrences of Stereotypy (*n*=186)**
Onset			**Onset**			**Onset**		
No change	*n*=49	*n*=63	No change	*n*=46	*n*=59	No change	*n*=82	*n*=92
Increase	*n*=60	*n*=39	Increase	*n*=45	*n*=30	Increase	*n*=66	*n*=49
Decrease	*n*=30	*n*=37	Decrease	*n*=22	*n*=24	Decrease	*n*=38	*n*=45
$X^2(2)=6.94$, $p<.05$ (more likely to be an increase at onset of occurrences)			**$X^2(2)=4.7$, $p=.10$**			**$X^2(2)=3.68$, $p=.16$**		
Offset			**Offset**			**Offset**		
No change	*n*=54	*n*=61	No change	*n*=47	*n*=63	No change	*n*=56	*n*=94
Increase	*n*=30	*n*=38	Increase	*n*=22	*n*=24	Increase	*n*=55	*n*=50
Decrease	*n*=55	*n*=40	Decrease	*n*=44	*n*=26	Decrease	*n*=75	*n*=42
$X^2(2)=3.74$, $p=.15$			**$X^2(2)=7.04$, $p<.05$ (more likely to be decrease at offset at offset of occurrences)**			**$X^2(2)=19.17$, $p<.001$ (more likely to be decrease at offset of occurrences)**		
Overall change in physiological arousal associated with stereotypy			**Overall change in physiological arousal associated with stereotypy**			**Overall change in physiological arousal associated with stereotypy**		
No change	*n*=43	*n*=54	No change	*n*=47	*n*=50	No change	*n*=72	*n*=66
Increase	*n*=52	*n*=41	Increase	*n*=27	*n*=27	Increase	*n*=55	*n*=69
Decrease	*n*=44	*n*=44	Decrease	*n*=39	*n*=36	Decrease	*n*=59	*n*=51
$X^2(2)=2.55$, $p=.28$			**$X^2(2)=0.21$, $p=.9$**			**$X^2(2)=2.42$, $p=.3$**		

	Participant 4			**Participant 5**	
	Occurrences of Stereotypy (*n*=143)	**Non-occurrences of Stereotypy (*n*=143)**		**Occurrences of Stereotypy (*n*=103)**	**Non-occurrences of Stereotypy (*n*=103)**
Onset			**Onset**		
No change	*n*=60	*n*=68	No change	*n*=35	*n*=62
Increase	*n*=32	*n*=40	Increase	*n*=40	*n*=25
Decrease	*n*=51	*n*=35	Decrease	*n*=28	*n*=16
$X^2(2)=4.37$, $p=.11$			**$X^2(2)=14.25$, $p=.001$ (more likely to be no change at onset of non-occurrences)**		
Offset			**Offset**		
No change	*n*=68	*n*=71	No change	*n*=46	*n*=59
Increase	*n*=37	*n*=38	Increase	*n*=26	*n*=20
Decrease	*n*=38	*n*=34	Decrease	*n*=31	*n*=24
$X^2(2)=0.30$, $p=.86$			**$X^2(2)=3.28$, $p=.19$**		
Overall change in physiological arousal associated with stereotypy			**Overall change in physiological arousal associated with stereotypy**		
No change	*n*=45	*n*=61	No change	*n*=40	*n*=42
Increase	*n*=43	*n*=48	Increase	*n*=35	*n*=33
Decrease	*n*=55	*n*=34	Decrease	*n*=28	*n*=28
$X^2(2)=7.65$, $p<.05$ (more likely to be an overall decrease associated with occurrences)			**$X^2(2)=0.11$, $p=.95$**		

Note. *n* refers to the number of occurrences of stereotypy or number of non-occurrences of stereotypy.

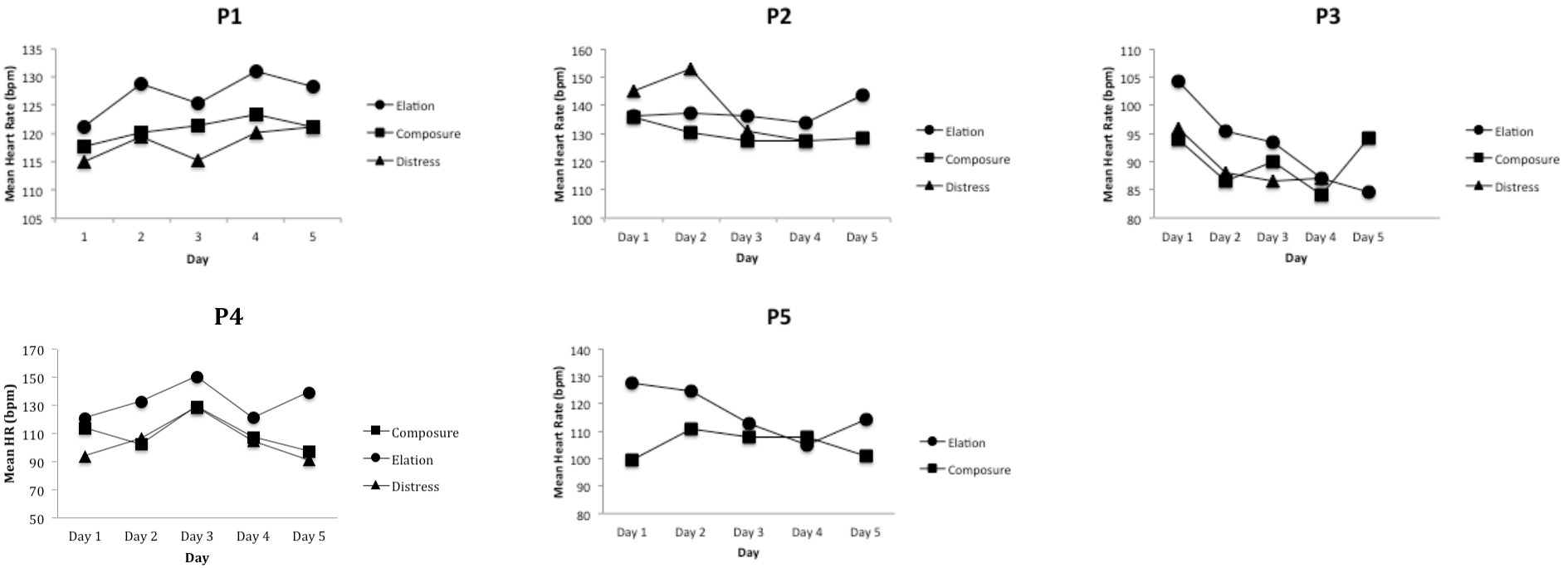

Figure 3. Mean heart rate patterns during occurrences of stereotypy associated with each mood state for Participant 1 (upper left panel), Participant 2 (upper middle panel), Participant 3 (upper right panel), Participant 4 (lower left panel), and Participant 5 (lower middle panel).

Participant 2's mean HR was found to be 130.77 bpm (*SD*= 14.72; range 76-194 bpm). The analysis of HR patterns surrounding stereotypy occurring during composure (see upper middle panel Table 11), elation (see upper middle panel Table 12), and distress (see upper right panel Table 13) failed to reveal any HR patterns that were associated significantly more frequently with occurrences of stereotypy that during non-occurrences of stereotypy. The analysis of HR patterns surrounding stereotypy (see upper right panel Table 14) yielded one significant difference between occurrences and non-occurrences; occurrences of stereotypy were significantly more likely to be followed by a decrease at offset than non-occurrences were. Next, mean HR during stereotypy occurring during each mood state for Participant 2 was analysed and is presented in the upper middle panel of Figure 3. Visual inspection suggested that HR was variable during occurrences for each mood state, although HR appeared to be consistently higher during elated occurrences than during those occurring during composure and distress. The exploration of this data using a one-way between-subjects ANOVA suggested that, under appropriate conditions, there would be a statistically significant difference in mean HR between stereotypy occurring during the different mood states, $F(2,796)=23.86$, $p=.001$. A Tukey post-hoc test revealed that HR was significantly higher, $p<.001$, during elation (M=136.4, SD=15.98) than during composure (M=128.92, SD=10.08) and significantly higher during distress (M=139.03, SD= 14.46) than composure, $p<.001$. There were no significant differences in HR during distress and elation.

Participant 3's mean HR was found to be 90.33 bpm (*SD*= 13.31; range 63-223 bpm). The analysis of HR patterns associated with stereotypy occurring during each of the mood states revealed no patterns that were significantly more likely to occur during incidents of stereotypy during distress (see upper right panel Table 13). However, it was observed that it was significantly more likely for HR increases or decreases to occur at the offset of occurrences of stereotypy during composure than it was for non-occurrences of

stereotypy (see upper right panel Table 11). Furthermore, the offset of occurrences of stereotypy observed during elation were significantly more frequently followed by HR decreases than was observed during non-occurrences (see lower left panel Table 12). The analysis of HR patterns co-occurring with all occurrences of stereotypy revealed that a decrease in HR post-offset was significantly more frequent with non-occurrences (similar to Participant 2; see lower left panel Table 14). Finally, Participant 3's mean level of HR during occurrences of stereotypy associated with composure, elation, and distress is depicted in the upper right panel of Figure 3. Visual analysis suggested HR was typically highest during elated occurrences and lower during occurrences associated with composure or distress. Exploration of this data using a one-way between-subjects ANOVA revealed that, were the assumptions of the ANOVA met, the differences observed through visual analysis would be highly statistically significant, $F(2,1416)=21.25$, $p=.001$. A Tukey post-hoc test revealed that HR was significantly higher during elation ($M=94.43$, $SD=13.74$) than during composure ($M= 89.19$, $SD=10.64$), $p<.001$, higher during elation than during distress ($M=89.08$, $SD=9.1$), and did not differ between distress and composure.

Participant 4's mean HR was found to be 118.5 bpm ($SD= 20.6$; range 69-194 bpm). For Participant 4, there were no HR patterns that were consistently associated with stereotypy during composure (see lower left panel Table 11), elation (see lower left panel Table 12), or distress (see lower right panel Table 13); the frequency of occurrence of the various HR patterns at onset, offset, and the change in physiological arousal as a result of stereotypy, did not deviate significantly from the frequency of those patterns during non-occurrences of stereotypy. The analysis of HR patterns across all occurrences of stereotypy (see lower middle panel Table 14) yielded one significant difference for Participant 4; an overall decrease in HR as a result of engagement in stereotypy was more frequently associated with occurrences than non-occurrences. The outcomes of the analysis of HR level during occurrences of stereotypy associated with all three mood states are presented

in the lower left panel of Figure 3. Visual inspection revealed HR was highest during occurrences of stereotypy during elation and lower during composure and distress. A one-way between-subjects ANOVA revealed that, if the assumptions of ANOVA were met, a statistically significant difference in mean HR between stereotypy occurring during different mood states, $F(2,11294)=433.79$, $p<.001$, would be observed. A Tukey post-hoc test revealed that HR was significantly higher during elation (M=136.11, SD=23.61) than during composure (M= 114.29, SD=17.51), $p<.001$, higher during elation than during distress (M=97.9, SD=10.19), and higher during composure than during distress, $p<.001$.

Participant 5's mean HR was found to be 109.7 bpm (SD= 17.5; range 60-193 bpm). The analysis of HR patterns associated with occurrences of stereotypy revealed two significant findings. First, as can be seen in the lower right panel of Table 11, occurrences of stereotypy associated with composure were significantly less frequently preceded by stable HR than non-occurrences were. Instead, occurrences of stereotypy during composure were more likely to be preceded by HR increases or decreases. Second, occurrences of stereotypy associated with elation were significantly more likely to result in a decrease in HR than non-occurrences were (see lower right panel Table 12). For Participant 5, the analysis of HR patterns associated with all occurrences of stereotypy indicated that non-occurrences were significantly more likely to be associated with "no change" in HR at onset than were occurrences of stereotypy (see lower right panel Table 14). Participant 5's level of HR during stereotypy was also analysed (see lower middle panel Figure 3). Visual inspection suggested that HR was higher during occurrences of stereotypy associated with elation than during occurrences associated with composure. As there were no occurrences of distressed stereotypy, an independent samples t-test was used to compare HR associated with stereotypy during composure and HR during stereotypy associated with elation. There was a statistically significant difference in mean HR between stereotypy occurring during the different mood states, $t(663.16)=9.27$, $p<.001$,

with mean HR significantly higher during elation (*M*=118.28, *SD*=22.86) than during composure (*M*=106.65, *SD*=12.9).

Discussion

The current study aimed to investigate the relationship between HR and motor stereotypy in individuals with autism. The results are suggestive of little relationship between these variables for four of the five participants who each presented with high frequency repetitive behaviours that functioned as automatic reinforcement. These findings are in contrast with the results of a number of studies conducted over the past five decades and have important implications for the further study of the association between physiological activity and stereotypy in this population. The findings regarding mean HR during various mood states may also make an important contribution to our understanding and knowledge of physiological functioning and PR to stressors in autism.

The results of the current study regarding the association, or lack thereof, between motor stereotypy and physiological activity are in contrast with a number of previous research studies in this area (Hutt et al., 1975; Lydon et al., 2013; Sroufe et al., 1973; Willemsen-Swinkels et al., 1998; Young & Clements, 1979). While previous studies have differed in the nature of the relationship observed between these variables (i.e., whether stereotypy is arousal-inducing or de-arousing), all have identified some correlation between the two variables. In the current study, there was little evidence to suggest that changes in HR functioned as an antecedent to, or reinforcing consequence of, the observed motor stereotypy for most of the participants. The analysis of all occurrences of stereotypy for each participant did yield a number of significant findings including: the greater likelihood of HR increases at onset of occurrences of stereotypy (Participant 1); the greater likelihood of a HR decrease at the offset of stereotypy (Participants 2 and 3), and the overall decrease in HR as a result of stereotypy (Participant 4). However, these patterns, while occurring significantly more frequently during occurrences of stereotypy than non-

occurrences, were not highly consistent within participants, and certainly not across participants as reported in previous studies (e.g., Lydon et al., 2013; Willemsen-Swinkels et al., 1998). Some of these patterns may be attributable to expected HR activity surrounding movement (i.e., a HR increase at the onset of movement, a decrease in HR following the cessation of movement) while others are less easily explained. For example, Participant 4's stereotypy was significantly more likely to result in an overall decrease in HR (i.e., decrease in HR from pre-stereotypy levels to post-stereotypy levels). In this case, it is possible that stereotypy functioned as an arousal reduction mechanism for this participant and this finding may inform direct intervention to reduce or eliminate stereotypy. For example, intervention focused on teaching the participant alternative appropriate arousal reduction techniques, such as relaxation skills or functional communication skills to request a low arousal environment, may result in a reduction in engagement in stereotypy.

While Willemsen-Swinkels and colleagues (1998) found that HR patterns associated with motor stereotypy differed according to the associated mood state, our analysis failed to replicate their results. It is possible that differences in research methodology (e.g., frequency of HR measurement), participant characteristics (e.g., degree of co-occurring intellectual disability, non-inclusion of participants with diagnoses other than autism in this study), and different methods of data analysis may have contributed to the inconsistencies between our findings and those of other similar research studies. However, the observation of inconsistent findings is not unique in research investigating physiological activity or functioning among persons with autism; Lydon and colleagues (in press/ Study 1: Chapter 2) reviewed studies assessing PR to stimuli among persons with autism and noted that the body of research was characterised by inconsistent findings even when highly similar research methodologies and samples were employed. Further research in this area is required to determine whether extraneous variables may explain the

relationship between physiological activity and stereotypy that was observed in previous research studies or whether there are characteristics of the individual or the target behaviours that predict whether this relationship is observed. Previous research examining physiological activity in autism has been suggestive of the presence of hypo- and hyper-aroused individuals with autism along with normally aroused individuals (Lydon et al., in press/ Study 1: Chapter 2). Physiological activity presents notably differently for each of these groups and it is possible that the relationship between stereotypy and physiological arousal may differ, or be present or absent, based on group membership.

The lack of a consistent association between physiological activity and stereotypy in the current study raises a number of questions and future research areas of interest for behaviour analysts. For example, research interest in the efficacy of antecedent exercise as an intervention for stereotypy among persons is growing (e.g., Morrison, Roscoe, & Atwell, 2011; Neely, Rispoli, Gerow, & Ninci, 2015), although the evidence base for this as an effective intervention is not yet conclusive (Kasner, Reid, & MacDonald, 2012). The employment of antecedent exercise interventions in a function-based manner first requires the demonstration that stereotyped behaviours serve to increase physiological arousal, a finding that has been observed in previous research studies (e.g., Lydon et al., 2013; Sroufe et al., 1973; Young & Clements, 1979) but not in the current study. Our results suggest that stereotypy may not be associated with physiological arousal for all individuals with autism and, importantly, that physical exercise may be unlikely to produce decrements in motor stereotypy among individuals with autism for whom the two variables appear unrelated, as in the current study. Further research that examines the mechanisms through which antecedent exercise produces its effects is necessary and may also serve to illuminate the relationship between physiological arousal and stereotypy.

Our findings also highlight the need for the further research and elucidation of automatic reinforcement processes. Scholars have previously noted that the concept of

automatic reinforcement is problematic and an identified behavioural function of this nature negates any usefulness for treatment development (Cunningham & Schreibman, 2008; LeBlanc et al., 2000; Vollmer, 1994). While previous research studies have suggested that changes in physiological arousal may act as a reinforcing consequence for motor stereotypy (e.g., Lydon et al., 2013; Sroufe et al., 1973; Willemsen-Swinkels et al., 1998; Young & Clements, 1979), the current study suggests that alterations in physiological arousal do not appear to be the source of automatic reinforcement in all cases of motor stereotypy, although it possibly contributed to engagement in stereotypy for Participant 4. Given the difficulties associated with the treatment of stereotyped behaviours in autism, and the negative consequences of engagement in such behaviour, there is a need for research that isolates and identifies the precise source and nature of the "automatic" reinforcement produced by these behaviours. Further research in this area may contribute to identifying more effective behavioural interventions related to differential functional outcomes.

Finally, our findings do call into question the utility of the addition of measures of physiological activity to functional assessments of stereotyped motor behaviours. Romanczyk and Gillis (2006) stated that the conceptual validity of such analysis is evident but called for investigation of their clinical validity. The current study offers clinicians an accessible and straightforward protocol for data collection and analysis but offers little support for the utility of such analyses for most individuals with autism and stereotyped behaviour. However, given the sizable body of prior research linking physiological activity or activation and motor stereotypy (e.g., Sroufe et al., 1973; Lydon et al., 2013; Lydon et al., 2015b/Study 2: Chapter 3; Willemsen-Swinkels et al., 1998; Young & Clements, 1979) along with the findings of the current study regarding Participant 4's stereotypy, such analyses must not be completely discounted. Instead, future research should endeavor to identify the most appropriate target behaviours, individuals, and methodologies for such

analyses.

An interesting finding emerging from the current study was the observation that, for all of the participants for whom occurrences of stereotypy during distress was observed, mean HR during distress was typically lower than the level of HR during elation. This is a surprising finding given that stress is typically associated with increased HR, as is happiness or elation (Pressman & Cohen, 2005). Further, for these participants, behavioural severity appeared more uniformly high (i.e., moderate or severe rather than mild) across occurrences of stereotypy associated with distress than across occurrences that were associated with composure or elation suggesting that the intensity or vigour of the behaviours may not explain the differences in HR observed across mood states. Previously, Lydon et al. (2013) reported that a participant in their study showed low levels of physiological activation during occurrences of self-injury and tantrum behaviours although the behaviours were evoked by a stressor. In the current study, indicators of participant distress included crying, tantrum behaviours, negative verbal comments, attempts to escape the situation, and negative facial expressions. With the exception of Participant 5, all participants were noted to be distressed on numerous occasions during the recording period in response to a variety of stimuli or apparently stressful contexts including the removal of preferred tangibles, the introduction of academic demand, the presence of noise or aversive auditory stimuli, known high-anxiety contexts, and the non-reinforcement of mands. The consistent observation of an overt distress response in the absence of a co-occurring physiological response for our participants warrants further consideration and research. Goodwin et al. (2006) previously observed under-responsivity to stressors among individuals diagnosed with autism. However, in Goodwin et al.'s study, participants with autism appeared hyper-aroused (i.e., had high resting HR) which was not the case in the current study. Jansen et al. (2006) alternatively suggested that a reduced stress response among persons with autism might be the result of years of chronic stress or physiological

activation that leads to the down-regulation of the central nervous system, a difficult hypothesis to confirm or refute. Alternatively, this finding could be indicative of blunted physiological responses to stress among persons with autism.

While research has focused predominantly on those who exhibit exaggerated, or increased, reactivity to stressors and the negative outcomes of this, there has been growing research interest in those who fail to respond physiologically to stressors and the implications of this in recent years (Ginty, 2013; Phillips, Ginty, & Hughes, 2013). Cardiovascular blunting has been defined as "an empirically demonstrable cardiovascular response pattern which is comparatively lower than that seen during typical states of homeostatic function during stress" (Phillips et al., 2013, p.2), and is associated with neural hypo-activation (Ginty, Gianaros, Derbyshire, Phillips, & Carroll, 2013). Among typically developing persons, physiological blunting has been associated with a myriad of negative outcomes including obesity (Phillips, Roseboom, Carroll, & de Rooij, 2012) and engagement in maladaptive activites such as smoking (al'Absi, Hatsukami, & Davis, 2005), alcohol and substance abuse (Lovallo, Dickensheets, Myers, Thomas, & Nixon, 2000), and antisocial behaviour (Ortiz & Raine, 2004). Blunted PR has also been linked to a number of psychological disorders including depression (Schwerdtfeger & Rosenkaimer, 2011), eating disorders (Ginty, Phillips, Higgs, Heaney, & Carroll, 2012), addictive disorders (Paris, Franco, Sodano, Frye, & Wulfert, 2010), and ADHD (Pesonen et al., 2011). Of perhaps primary relevance to persons with autism, PR has also been associated with engagement in challenging behaviour (Bauer, et al. 2002), patterns of social interaction (Kagan et al., 1987), and emotional reactivity (Fox, 1989).

A number of explanations for physiological blunting have been put forward; these include the suggestion that blunted PR is an outcome of lower awareness or reduced perception of the stressor, that physiological blunting may be an outcome of early life stress, that physiological blunting may be an outcome of physiological dysfunction in the

ability to respond to stress or may be the result of dysregulation in the areas of the brain responsible for motivation (Phillips et al., 2013). One factor that may mediate the relationship between autism and physiological blunting is cognitive ability. Research has shown that reduced physiological responsivity is associated with lower cognitive ability (Ginty, Phillips, Roseboom, Carroll, & Derooij, 2012). The majority of participants in the current study were diagnosed with a co-occurring intellectual disability, or presented with below-average cognitive ability. Such impairments in cognitive ability may impact upon physiological responsivity by reducing awareness or understanding of the stressor (Phillips et al., 2013). A number of studies have identified differences in baseline physiological functioning by level of cognitive functioning (e.g., Putnam et al., 2015; Tordjman et al., 2014) and future research examining PR among low- and high-functioning individuals with autism may provide us with greater understanding of the impact of cognitive ability on PR in this population. It is important to note, however, that physiological blunting may not be a wholly negative or undesirable reaction; Phillips and colleagues (2013) have suggested that physiological blunting in response to stress, while associated with negative health and behavioural outcomes, may confer some advantages during stressful situations. For example, Phillips et al. propose that this physiological non-response may be associated with better short-term coping, stress management, or communication and social interaction. The investigation of the possible benefits of physiological non-response to stressors among persons with autism would be of much interest.

In the current study, the lack of division between HR during composure and distress, and the corresponding gap between HR during elation and distress, possibly reflects physiological blunting to stressors among the included participants. Heretofore, the majority of research on physiological blunting in response to stress has been conducted by measuring PR to laboratory stressor tasks, a context wholly different from that of the current study. However, we suggest that, given the findings of the current study and those

of Lydon et al. (2013), that the phenomenon of physiological blunting, and the implications of this, be investigated further among persons with autism. Currently, the body of research on PR to stressors and other stimuli among persons with autism (see Lydon et al., in press/Study 1: Chapter 2 for a review) suggests that both physiological under- and over-reactivity are relatively common among persons with autism as compared to typically developing controls. Studies in this area have also frequently noted the presence of non-responders, or high levels of variability in responsivity, among members of the autism group (Lydon et al., in press/Study 1: Chapter 2). Such findings suggest that physiological blunting to stressors may not be present for all individuals in autism but is potentially relatively common. However, studies in this area have been criticised for poor participant characterisation (Lydon et al., in press/Study 1: Chapter 2), thus research exploring the cognitive, behavioural, and physiological profiles of physiologically under- or overly-responsive individuals with autism may further our understanding of the disorders and of atypical behaviours.

The current study had a number of limitations. Key among these was the reliance upon convenience sampling; we recruited from a broad sample of children and adolescents who engaged in motor stereotypy that was reported to interfere with their academic learning or social opportunities. Different results may have been obtained had we sought participants for whom motor stereotypy appeared overtly to be associated with physiological arousal (i.e., increased respiration rate, flushing etc.) or for whom physiological arousal was hypothesised to play a role in their stereotypy. Future researchers may wish to recruit individuals with autism for whom physiological arousal is hypothesised to be related to motor stereotypy in some way. The process of forming such hypotheses may not be straightforward but could be accomplished by considering or assessing the impact of other forms of stimulation such as visual, auditory, or proprioceptive input on the behaviours. Previous research studies have successfully

demonstrated the formation of hypotheses surrounding the sensory stimulation yielded by stereotypy and the testing of these (e.g., Tang, Patterson, & Kennedy, 2003). The examination of the frequency with which stereotypy occurs during low-arousal (e.g., rest periods, relaxation breaks) and high-arousal periods (e.g., physical exercise, situations known to be stressful for the individual) may also yield information suitable for forming a hypothesis about the relationship between stereotypy and physiological arousal prior to the examination of this relationship in the manner reported in this study.

A further limitation of the current study was the reliance upon an indirect functional assessment tool to determine that participants' motor stereotypy was primarily non-socially mediated. While the QABF has been demonstrated to be a valid measure of behavioural function among persons with autism (Healy et al., 2013), it is possible that an experimental functional analysis would have yielded different information regarding the function of participants' behaviour. Further, it was not possible to discern the function of individual occurrences of motor stereotypy during the recording period. Future research may aim to examine the utility of measures of physiological activity during experimental functional analyses. Such methodologies would allow for the more accurate assessment of behavioural function. The use of a functional analysis methodology would also allow for the examination of physiological patterns associated with specific behavioural functions. For example, in the current study, all participants' motor stereotypy was suggested to serve multiple functions by the QABF. It is possible that had the function of each occurrence been known that the analysis of HR patterns by behavioural function might have yielded different outcomes to those observed in the current study. Future research should investigate this possibility.

The method of data analysis in the current study may be critiqued also. Visual inspection of the participants' data suggested that HR level during distress was notably lower than HR during elation, and similar to HR during composure. While the use of

statistical analysis for data originating from single-subject research designs can only ever be tentative (Baer, 1977; Michael, 1974), the use of one-way between subject ANOVAs in the present study helped to corroborate what was apparent from visual inspection. The data from each ANOVA arose from single individuals, and so violated the assumption of true independence of observations. However, ANOVA is somewhat robust against violations of assumptions, and in the present study there was the opportunity to corroborate any tentative finding by deploying the procedure in an exploratory manner across four separate participants. Doing so revealed distinct and recurring patterns of statistical significance in three of the four participants, consistent with the conclusions drawn from visual inspection. The fact that the significance and direction of these effects were the same across three participants supports the conclusion that the effects seen were in fact meaningful, rather than solely being errors that arise from violations of ANOVA assumptions.

Finally, the use of a single measure of physiological activity, HR, in the current study may also be considered a limitation. HR is highly labile and is influenced by a myriad of factors including movement, body positioning, respiration rate, environmental conditions, and mental or emotional state (Palatini, 2009). While previous research studies have successfully identified a relationship between HR and stereotypy using only a measure of HR (e.g., Lydon et al., 2013; Sroufe et al., 1973; Willemsen-Swinkels et al., 1998; Young & Clements, 1979), scholars have recommended the usage of multiple measures of physiological activity during research studies of this nature (Cohen et al., 2011; Romanczyk & Matthews, 1998). Researchers have successfully employed other measures of physiological activity in the study of challenging behaviour such as EDA (e.g., Romanczyk & Matthews, 1998) and cortisol (Gabriels et al., 2013; Lydon et al., 2015b/Study 2: Chapter 3). Future research studies in this area may wish to employ multiple measures of physiological activity in order to better our understanding of how physiological activity and challenging behaviour may be related among persons with

autism.

The aims of Study 4 (Chapter 5) were to improve upon the methodology of the current study in a number of ways that may contribute to an improved ability to detect any relation that may exist between physiological activity and engagement in repetitive, stereotyped behaviours among persons with ASD. Study 4 (Chapter 5) thus employed two measures of physiological activity, HR and the less labile EDA, in the context of a more systematic behavioural assessment, an experimental functional analysis, for individuals whose stereotyped, repetitive behaviour was at least partly maintained by automatic reinforcement or had proved resistant to prior intervention. The findings of the current study, regarding possible physiological blunting to stressors among children and adolescents with autism, were also further explored in the chapter that follows.

Chapter 5: Study 4

An Examination of the Physiological Correlates of Challenging Behaviour in Autism Spectrum Disorders during Functional Analysis

Abstract

A growing body of theoretical and empirical literature is suggestive of a relationship between physiological activity and engagement in challenging behaviour among persons with autism. Experimental functional analysis has previously been criticised for failing to consider this potential relation between physiological activity and challenging behaviour. The current study sought to present a model that may have utility for practitioners in conducting functional analysis, while capturing measures of heart rate and electrodermal activity, and for analysing the resultant data. Six children and adolescents diagnosed with autism who engaged in high rates of repetitive, stereotyped behaviours participated in this study. The results revealed little association between physiological activity and engagement in repetitive behaviours among these participants. Participants' physiological responses to stress were also investigated but physiological blunting, as suggested for a number of participants in Study 3, was only indicated for one of the four participants for whom distress was observed during the functional analysis. The implications of these findings for the clinical utility of psychophysiological assessment of challenging behaviour and our understanding of stress responsivity among persons with autism are discussed. Guidance for future research on automatic reinforcement processes and the psychophysiology of ASD is also provided.

Functional analysis is widely considered to be the most effective and useful tool in the area of ABA (Delfs & Campbell, 2010; Hanley, 2015; Mace, 1994). Comprising the manipulation of establishing operations and behavioural consequents, functional analysis procedures allow for the determination of environmental variables that contribute to engagement in challenging behaviour (Hanley et al., 2003; Iwata et al., 1994b). Functional analysis procedures have been widely examined in the research literature with over 400 studies reporting their usage (for reviews, see: Beavers, Iwata, & Lerman, 2013; Hanley et al., 2003). Importantly, the emergence of functional analysis has contributed to the increased use of reinforcement-based procedures in the treatment of challenging behaviour and a decreased use of punishment-based interventions (Pelios, Morren, Tesch, & Axelrod, 1999). Further, a wealth of research has shown that behavioural interventions based on a prior functional analysis are more effective than those developed without the assistance of a functional analysis (Campbell, 2003; Heyvaert et al., 2014; Kahng et al., 2002).

However, in spite of these successes, functional analyses are not without limitations; these procedures are time consuming, require significant expertise, and are unsuitable for use with certain challenging behaviours such as low rate behaviours or physically harmful behaviours, and may reinforce challenging behaviour (Iwata et al., 2000; Najdowski, Wallace, Ellsworth, MacAleese, & Cleveland, 2008; Tincani, Castrogiavanni, & Axelrod, 2009). It has also been suggested that the conditions included in a traditional functional analysis may be too narrow to provide an understanding of behaviours maintained by more complex sources of reinforcement; Carr (1994) highlighted that current functional analysis techniques are not useful in identifying individuals whose challenging behaviour is maintained by biological reinforcement. Further, in a small percentage of cases, functional analyses fail to identify the source of reinforcement responsible for the maintenance of the challenging behaviour under investigation (e.g., Beavers et al., 2013; Hanley et al., 2003; Iwata et al., 1994a; Kurtz et al., 2003); Kurtz and

colleagues (2003) found that functional analysis results were undifferentiated for 37.9% of their participants with SIB (*n*=29). However, a review by Hanley and colleagues (2003) found that, across 536 functional analysis datasets, only 22 cases (4.1%), primarily cases of SIB, resulted in undifferentiated functional analysis outcomes. These limitations of traditional functional analysis methodologies likely contribute to the poor rates of functional analysis usage reported by clinicians (e.g., Desrochers, Hile, & Williams-Mosely, 1997; Ellingson, Miltenberger, & Long, 1999; Weber, Killu, Derby, & Barretto, 2005).

A variety of adaptations to traditional functional analysis have been developed in order to remedy the perceived limitations of the original model (for a review of these, see: Lydon, Healy, O'Reilly, & Lang, 2012). However, one adaptation that has received limited research attention is that of a functional analysis that incorporates measures of physiological activity in order to assess potential physiological reinforcers for challenging behaviour. A potential link between physiological activity and challenging behaviour has been long and widely hypothesised (e.g., Guess & Carr, 1991; Hutt et al., 1964; Hutt & Hutt, 1968; Lydon et al., 2013; Romanczyk, 1986; Sugarman et al., 2014). Such theories have prompted experimental investigations of the relationship between ANS (for a review of these, see: Chapter 1; Cohen et al., 2011) or LHPA axis functioning (e.g., Gabriels et al., 2013; Hall et al., 2008; Lydon et al., 2015b/Study 2: Chapter 3; Symons et al., 2003; Symons et al., 2011; Verhoeven et al., 1999) and engagement in challenging behaviours such as SIB and stereotypy among persons with developmental disabilities. Romanczyk and Matthews (1998) proposed that physiological events may be "antecedents, determinants, concomitants, or the results of behavior" (p. 117) while Baker and colleagues (2002) also described the potential for physiological events to function as setting events for problem behaviour. These authors highlight the potential utility of psychophysiological measurement for contributing to the understanding and prediction of

multiply controlled or automatically reinforced challenging behaviour. Romanczyk and Matthews (1998) called for single-subject, empirical investigations of the clinical usefulness of psychophysiological assessments for individuals with autism. Similarly, Baker et al. (2002) have called for the consideration of both physiological and environmental variables during the functional analysis of challenging behaviours.

In spite of this consensus regarding the need to consider physiological activity during the functional assessment or analysis of complex cases of challenging behaviour, little research has examined the relationship between these variables in such contexts. Of primary relevance to the current study are a small number of research studies that employed measures of physiological activity during experimental analyses of behaviour. Barrera and colleagues (2007) conducted functional analyses including HR measures for three adults with developmental disabilities who engaged in chronic, severe SIB. They identified a HR pattern associated with the SIB of all three participants. This HR pattern consisted of increased HR prior to, or during, SIB followed by a significant decrease in HR post-SIB and was not impacted upon by the functional analysis conditions. The authors theorised that elevated physiological arousal served as an antecedent for SIB and that engagement in SIB was negatively reinforced by decreases in physiological arousal. Several years later, Hall and colleagues (2013) conducted a similar study to assess whether physiological activity contributed to engagement in severe skin picking by a child diagnosed with prader-willi syndrome. The results of this analysis indicated that HR was consistently higher during skin picking than was typical for that individual leading the authors to suggest that the behaviour was automatically reinforced by the increases in arousal that it produced. Finally, Moskowitz and colleagues (2013) examined the relationship between behavioural indicators of anxiety, challenging behaviour, and HR for three children diagnosed with autism. During the study, the authors systematically manipulated variables to produce either “high-anxiety” or “low-anxiety” environmental

conditions. The authors found that both HR and challenging behaviour were consistently higher during the “high-anxiety” condition for all participants. The authors used these findings to highlight the potential impact of internal states on challenging behaviour and the potential role of anxiety and its physiological concomitants as a setting event or a discriminative stimulus for engagement in challenging behaviour. Collectively, these studies indicate the promising feasibility and clinical utility of functional analyses incorporating measures of physiological activity.

The previous studies in this research program were suggestive of some relation between physiological activity and engagement in repetitive, stereotyped behaviours for some individuals with autism. However, when present, the nature of this association appeared to vary among participants with autism; Study 2 (Lydon et al., 2015b/Chapter 3) tentatively suggested that stereotyped behaviour was an indicator of elevated physiological activation, but did not function to mitigate physiological activity, while Study 3 (Lydon, Healy, Mulhern, & Hughes, 2015c/Chapter 4) suggested that, for one participant, stereotyped motor behaviours may have functioned to reduce physiological arousal. The aim of the current study was to expand upon this extant research base by examining the clinical utility of a practitioner-friendly model of functional analysis incorporating two physiological measures for six individuals diagnosed with ASD who engaged in severe repetitive, stereotyped behaviours and further assessing the association between physiological activity and repetitive, stereotyped behaviour in this population. Participants were selected as their repetitive, stereotyped behaviours were identified via indirect assessment to be either automatically reinforced or multiply controlled, or had been resistant to prior function-based behavioural intervention. Thus, this study attempted to determine whether the analysis of co-occurring physiological activity could yield a greater understanding and suggestions for treatment for these children and adolescents.

Method

Ethical Approval, Participants, Target Behaviours, and Setting

The study was approved by an ethical research review board at the National University of Ireland Galway. Consent was sought first from parents or caregivers, and then participants where possible.

Six individuals diagnosed with ASD were recruited to participate in this study. None of these participants had taken part in Study Three. All participants engaged in high rates of repetitive, stereotyped behaviours that were reported to interfere with their educational attainment and their social inclusion. For inclusion in the study, all participants had to display stereotyped behaviour that was either automatically reinforced, as indicated by the QABF (Matson & Vollmer, 1995), or repetitive, stereotyped behaviour that had proved resistant to prior function-based behavioural intervention.

Participant 1 was a 16:4 year-old male diagnosed with autism, a moderate intellectual disability, and obsessive compulsive disorder. His weight was 45 kg and his height was 162.5 cm. Participant 1 was receiving 5 mg of olanzapine twice daily throughout the study; this medication was prescribed with the aim of reducing the challenging behaviours that were investigated in the current study. Participant 1 had attended a special school that delivered ABA educational interventions from the age of 4. He was verbal but rarely engaged in spontaneous communication with others. Typically, verbal communication was limited to one word utterances that functioned as mands. Participant 1 engaged in three classes of repetitive, stereotyped behaviours that were of concern to his parents and school staff. These included multiple topographies of repetitive SIB, motor stereotypy, and repetitive “fixing” behaviours. The topographies of behaviour comprised by each of these classes are presented in Table 15. Each class of challenging behaviour had proved resistant to prior function-based behavioural interventions including DRO, planned ignoring, time-out, DRI, noncontingent reinforcement, and DRO and

noncontingent reinforcement in combination. While Participant 1's stereotyped motor behaviour had been present at diagnosis, repetitive SIB had emerged six years prior to the current assessment, and the repetitive fixing behaviours had first been observed two years prior to the current assessment. Near-constant engagement in SIB and motor stereotypy led to frequent tissue damage, interfered greatly with his ability to engage with educational material or social interactions, and created difficulties around transitions and traveling that resulted in the severe restriction of Participant 1's environment. The results of three QABFS completed by the school's director of behavioural services, indicated that each class of behaviour was multiply controlled with both non-social and social functions. QABF data for each participant in this study are presented in Appendix B.

Table 15

Topographies and Operational Definitions for the Repetitive, Stereotyped Motor Movements of all Participants

Topography	Operational Definition	P1	P2	P3	P4	P5	P6
Repetitive Motor Movements							
Body banging	bringing fist(s), elbow(s), or open palm(s) down forcefully onto a surface, the individual's own body, or another's body			✓			✓
Body bouncing	moving up and down quickly while standing, seated, or moving.			✓			
Body circling	rotating torso in smooth circular motion						✓
Body contortions	twisting torso or moving shoulders up and down	✓					
Body rocking	tilting body backwards and forwards in fast or slow motion		✓		✓	✓	✓
Body swaying	shifting torso from one side to another multiple times			✓			
Hand clapping	hitting hands together, while flat or in fists		✓			✓	✓
Hand flapping	moving one or both hands up and down quickly	✓	✓	✓	✓	✓	✓
Hand in mouth	inserting hand with flat palm into mouth and removing it quickly						✓
Hand patting	lightly touching skin, body parts, surfaces, or objects with open palm of one or more objects		✓	✓	✓		
Hand pressing	pushing palms together forcibly			✓	✓		
Hands to face	pressing the back of both hands, or clenched hands, to face near mouth area		✓		✓	✓	
Head bobbing	moving head up and down, or from side to side, quickly without communicative intent			✓		✓	
Jumping	leaping of the ground multiple times in quick succession when walking, running, or stationary		✓	✓	✓		✓
Limb swinging	moving limbs back and forth, or up and down, quickly			✓	✓	✓	
Mouthing	placing object or finger(s) in mouth and closing lips or teeth around object lightly		✓				
Finger movements	clenching fingers together, wiggling fingers, tapping or flicking nose or other objects with fingers	✓		✓	✓	✓	✓
Rubbing	brushing hands together or rubbing hands along skin, hair, body parts, surfaces, or objects with open palm(s) or fingers		✓	✓	✓	✓	
Shrugging	moving shoulders up and down quickly	✓			✓		
Tapping	repeatedly touching head or forehead with finger(s) or objects					✓	
Repetitive "fixing" behaviours	repetitive action to re-order/alter position of/move/change materials, furniture, or items in the environment	✓					
Stereotyped object manipulation	• holding object in hand or fingers and tapping object on body part, surface, or other object • transferring object back and forth from one hand to the other hand multiple times in quick succession • picking up object and dropping it quickly into other hand or onto surface before picking it up again and repeating • twisting object between two hands • Squeezing objects in hand		✓				
Repetitive Self-injurious Behaviours (Participants 1 and 4)							
Self-biting	inserting one or more fingers into mouth and closing teeth on finger(s)	✓					
Elbow digging	pressing elbow(s) into side of torso or hips with force	✓					
Self-hitting	striking legs with fists or open palms	✓					
Self-scratching	scraping skin with fingernails	✓					
Self-pinching	gripping a section of skin tightly between finger(s) and thumb	✓					
Face-rubbing	rubbing face forcefully with closed fists	✓					
Hair pulling	pinching a section of hair between fingers and tugging	✓					
Skin and nail picking	pulling or digging at skin on fingers, pulling under fingernails with other fingernail or object, biting and pulling skin on fingers				✓		

Participant 2 was a 12:2 year-old male diagnosed with autism and a severe to profound intellectual disability. At the time of participation, he was undergoing psychological evaluation for a possible diagnosis of a co-occurring mood disorder. He was diagnosed with irritable bowel syndrome and was also under consideration for a possible diagnosis of temporal lobe epilepsy. His weight was 49.4 kg and his height was 160.2 cm. Participant 2 had been diagnosed with autism at the age of 2 year 10 months. He had attended a special school for children with autism that delivered ABA educational interventions for a four-year period. However, at the time of assessment he resided full-time in a residential service for children and adolescents with complex needs and severe challenging behaviour. He received 20 hours of tuition weekly from a Board Certified Behaviour Analyst® at his residential centre. Participant 2 engaged in numerous forms of severe challenging behaviour. These included SIB in the form of scratching, head banging, head kneeing, self slapping, body slamming, and self-biting. Participant 2 frequently required medical attention following engagement in these behaviours. SIB often co-occurred with aggressive behaviours towards other children and adults in his environment. Aggressive behaviours included scratching, slapping, biting, pinching, hitting, and kicking. These behaviours had previously resulted in staff injuries that required emergency medical attention and treatment. Participant 2 engaged in pica and coprophagy and had required medical attention in the past due to the ingestion of harmful materials. Participant 2 also engaged in problematic sexual behaviours including public masturbation and the exposure of his genitalia in public areas. Participant 2's participation in the current study, however, was due to his engagement in excessive stereotyped motor behaviour. Participant 2 engaged in near constant repetitive behaviour and was noted to become highly distressed if objects were not available with which he could engage in stereotypy. The topographies of Participant 2's stereotypy are outlined in Table 15. Participant 2's SIB and aggressive behaviours were excluded from analysis, and their occurrence was not consequated during

the functional analysis, due to their high severity and the possibility that even one occurrence of these behaviours could result in serious physical injury to Participant 2 or those in the experimental environment. A QABF completed by Participant 2's key tutor indicated that Participant 2's stereotyped behaviours were multiply controlled, sometimes serving as an indicator of physical discomfort and also sensitive to sources of negative reinforcement, automatic reinforcement, and positive reinforcement in the form of access to tangibles. There was no record of previous intervention for these behaviours. However, a behaviour support plan was in place throughout the assessment period for Participant 2's SIB and aggressive behaviours. The low arousal approach (see McDonnell et al., 2015) was the key strategy used to guide staff in the management of all challenging behaviours, as physiological arousal was hypothesised to play a role in their occurrence.

Participant 3 was a 7:10 year-old male diagnosed with autism. His height was 129.5 cm and his weight was 28.6 kg. Participant 3 was not receiving any psychotropic medication during the study. Participant 3 was diagnosed with autism at the age of 2 years and 3 months. He had received 20 hours of ABA educational intervention weekly for two years following his diagnosis. At the time of the study, he was attending an autism-specific special school that adhered to an eclectic model of intervention and received eight hours of ABA home tuition weekly. Participant 3 was verbal but rarely engaged in spontaneous verbal communication other than manding for preferred items. He engaged in near-constant vocal and motor stereotypy. Topographies of motor stereotypy that were measured during the current study are presented in Table 15. The results of a QABF completed by Participant 3's ABA home tutor indicated that these behaviours were multiply controlled, sensitive to sources of both automatic reinforcement and negative reinforcement and also serving as an indicator of physical discomfort on occasion. These behaviours were reported to be a significant impediment to engagement and learning by Participant 3's parents and ABA instructor. At the time of the study, an inhibitory stimulus

control intervention was being used in Participant 3's ABA home tuition sessions but was not considered to have resulted in a clinically significant reduction in stereotyped behaviours. This intervention was discontinued for the duration of the experimental analysis.

Participant 4 was an 11:10 year-old male diagnosed with autism and a mild intellectual disability. His height was 147 cm and his weight was 39.5 kg. Participant 4 was not receiving any medication during the study. He had received an ABA educational intervention from the age of 2 years and 6 months until 7 years of age. He was currently attending a special school for children with autism that adhered to an eclectic model of intervention. Participant 4 was verbal but rarely engaged in spontaneous communication with those in his environment. Verbal communication was typically comprised of tacts relating to preferred objects. Participant 4 was referred to the experimental team as he engaged in high rates of motor stereotypy and repetitive SIB in the form of skin picking (see Table 15 for more information on topographies). The results of QABFs completed by the school's director of behavioural services indicated that both of these classes of behaviour (repetitive SIB and repetitive motor stereotypy) were maintained solely by automatic reinforcement. Participant 4 also engaged in a number of other challenging behaviours including pica, bruxism, property destruction, and aggressive behaviours including biting, hitting, scratching, and kicking. These challenging behaviours were not consequated during the functional analysis of his stereotyped motor behaviour.

Participant 5 was a 9:1 year-old male diagnosed with moderate to severe autism, as per the CARS, at the age of 2 years and 3 months. He was also diagnosed with a co-occurring severe intellectual disability as per the Griffiths Mental Development Scales. His height was 136 cm and his weight was 34.5 kg. Participant 5 was attending the same special school for children with autism that adhered to an eclectic model of intervention as Participant 4. Participant 5 was non-verbal and communicated using a speech-generating

device. Communication consisted solely of mands for preferred items or activities. Participant 5 was selected to participate in the study due to his engagement in high rates of motor stereotypy that were reported to greatly impede his learning and engagement with learning tasks. The topographies of motor stereotypy in which he engaged are presented in Table 15. The results of a QABF completed by the school's director of behavioural services indicated that Participant 4's stereotyped behaviour was maintained solely by sources of automatic reinforcement. These behaviours had not been targeted for intervention previously.

Participant 6 was a 12:8 year-old male who had been diagnosed with autism and moderate to severe intellectual disability at the age of two years. His height was 156 cm and his weight was 40.3 kg. Participant 6 had received four years of ABA educational intervention following his diagnosis and was currently attending the same special school for children with autism as Participants 4 and 5. Participant 6 was non-verbal and communicated using the Picture Exchange Communication System®. Participant 6 engaged in a number of challenging behaviours including pica, elopement, aggression in the form of grabbing or pinching, and high rates of severe and vigorous motor stereotypy. The topographies of motor stereotypy that he exhibited which were measured in the current study are presented in Table 15. The results of a QABF completed by the school's director of behavioural services indicated that Participant 6's stereotyped motor behaviours were maintained solely by automatic reinforcement. These behaviours had not been subject to previous intervention.

For Participant 1, data collection was conducted at his school, a special school for children with autism that delivered ABA educational interventions. For Participant 2, data collection occurred at the residential centre where he resided and where he received ABA tuition on weekdays. Data collection for Participant 3 occurred in his home, in the room

where he typically received his ABA home tuition. Data collection for Participants 4, 5 and 6 occurred in a room at their special school.

Behavioural Recording

All functional analyses were video recorded by participants' school or service. The iPad® used to record sessions was synced with the time on the HR monitor and EDA monitors to ensure correspondence between the three and to allow for the accurate comparison of behavioural and physiological data. Raters subsequently used the videos to record the frequency of target behaviours that occurred during the functional analysis. Raters assessed the frequency of target behaviours using 10 second partial interval recording for each five minute condition. Data were also collected on the time of onset and offset of target behaviours, the class of target behaviour observed (for Participants 1 and 4 who engaged in both repetitive SIB and motor stereotypy, and also repetitive fixing behaviours for Participant 1), behavioural intensity, and the participant's mood state during the occurrence of the behaviour. Target behaviours were rated as mild, moderate, or severe according to the definitions presented in Study 3 (Lydon et al., 2015c/Chapter 4). Participant mood state was recorded as composure, distress, or elation as per the protocol described in Willemsen-Swinkels and colleagues (1998) and Study 3 (Lydon et al., 2015c/Chapter 4).

An interobserver viewed a minimum of two of the four experimental sessions for each participant. Interobserver agreement was assessed across raters' partial interval recording and across the four additional categories of behavioural recording (i.e., onset time, offset time, behavioural severity, and mood state) for Participants 2-6, and the five categories of behavioural recording for Participants 1 and 4 (i.e., class of behaviour, onset time, offset time, behavioural severity, and mood state). For partial interval recording, raters were considered to agree if they had both noted either the presence or absence of the target behaviour during a recording interval. The data recorded for the time of onset and

time of offset for each behaviour were considered to agree if the recording differed by two seconds or less, but the raters engaged in additional review of the videos to reach agreement on an exact time of onset or offset in all cases in which there was discrepancy in the time of onset or offset. For the other categories, raters were considered to agree if the information recorded matched, and to disagree if the information recorded differed. All disagreements were resolved through discussion by raters and additional review of the video recordings. For Participant 1, interobserver agreement was assessed across all four functional analysis sessions. Interobserver agreement was found to be 93.5% (73.3%-100%) for partial interval recording and 81.5% (range 66.9-96%) for behavioural recording. For Participant 2, interobserver agreement across two experimental sessions was found to be 97.3% (range 83.3%-100%) for partial interval recording and 96% for behavioural recording. For Participant 3, interobserver agreement across two experimental sessions was found to be 97.3% (range 86.7-100%) for partial interval recording and 93.1% for behavioural recording. For Participant 4, interobserver agreement was found to be 91.3% (range 73.3-100%) for partial interval recording and 83.1% for behaviour recording across the two experimental sessions reviewed by a second rater. For Participant 5, interobserver agreement across two experimental sessions was found to be 96.3% (range 90-100%) for partial interval recording and 96.2% for behavioural recording. For Participant 6, interobserver agreement across two experimental sessions was found to be 94.4% for behavioural recording and 99% (93.3%-100%) for partial interval recording. All disagreements were resolved through discussion by raters and additional review of the video recordings.

Physiological Recording

The use of multiple measures of physiological activity in research such as the current study has been recommended (Cohen et al., 2011; Lydon et al., 2015c/Study 3: Chapter 4). For this study, data were collected on HR and EDA during each functional

analysis. HR data were recorded using the Polar RS800cx (Polar Electro OY, Kempele, Finland) in the manner described previously in Study 3 (Lydon et al., 2015c/Chapter 4).

EDA was also measured in this study using Affectiva's Q-sensor (Affectiva, MA, USA). This lightweight device captures a measure of EDA, expressed in microSiemens (μS), from the wrist or the ankle eight times per second. The Q-sensor has previously been used with individuals diagnosed with developmental disabilities including autism (Gay, Leijdekkers, Agcanas, Wong, & Wu, 2013; Noordzij, Scholten, & Laroy-Noordzij, 2012; Mower, Black, Flores, Williams, & Narayanan, 2011). In the current study, all participants wore the Q-sensor on their ankle; the recording of EDA from the ankle using the Q-sensor has been demonstrated to produce data comparable to the recording of EDA from the wrist using the device (M. Goodwin, personal communication, June 17, 2013). Data on Participant 4's EDA during the brief functional analysis were not available due to the malfunctioning of the Q-sensor during data collection.

Procedure and Experimental Design

For each participant, the current study began with a two-day habituation period intended to acclimatise the participants to wearing the HR and EDA monitoring devices. As in Study 3 (Lydon et al., 2015c/Chapter 4), operational definitions for the topographies of stereotypy exhibited by each participant, presented in Table 15, were developed by the researcher during the habituation phase for use by the observer(s) during data collection. During this period, a multiple stimuli without replacement preference assessment (DeLeon & Iwata, 1996) was also conducted to identify preferred items for use during the subsequent functional analysis.

A brief functional analysis was subsequently conducted for all participants (Derby et al., 1992; Lydon et al., 2012; Northup et al., 1991) using a multi-element design. Prior to each functional analysis, participants were fitted with the HR and EDA monitors and the functioning of each of these was checked between conditions. For Participant 1, his class

teacher, a masters student in ABA, conducted all functional analysis sessions. For Participant 2, his ABA tutor, a Board Certified Behaviour Analyst®, conducted all functional analysis sessions. For Participant 3, his ABA tutor, a masters student in ABA, conducted the functional analysis sessions. For Participants 4, 5 and 6, their school's director of behavioural services, a Board Certified Behaviour Analyst®, conducted the functional analysis sessions. All functional analyses were also conducted in the presence of an independent Board Certified Behaviour Analyst®, a member of the research team.

For each participant, experimental conditions included attention, escape, alone, free play, and tangible and were of five minutes duration. For each participant, the brief functional analysis was carried out across one day. Four repetitions of each condition were conducted for all participants excepting Participant 2. Termination criteria were specified in advance. These included the termination of a condition if the stereotyped behaviour under study manifested into repetitive SIB that resulted in apparent visible tissue damage. Further, it was specified that a condition would be terminated if a participant began to engage in aggressive behaviour towards those present during the analysis. The functional analysis of Participant 2's stereotyped motor behaviour was terminated after three sessions due to an escalation in his aggressive behaviours that was considered unacceptably dangerous for those involved in the analysis. For Participant 1, an additional condition that examined rates of challenging behaviours during transitions was also conducted. The order of conditions was randomised throughout the functional analysis by drawing the condition names from a pot prior to each sequence. There was a minimum of five minutes separating the conclusion of one condition and the presentation of the next condition. The content of these conditions was based upon Iwata et al.'s (1994b) protocol and is described below.

Attention condition. Throughout this condition, the participant was given free access to a range of materials and directed to engage with them. The experimenter sat in a chair, away from the participant, and engaged with a magazine or book. Physical and

verbal attention, in the form of statements of concern and displeasure, were given to the participant for ten seconds contingent on the occurrence of any of the target behaviours. This condition was intended to assess whether the target behaviours were maintained by access to positive reinforcement in the form of attention from others.

Escape condition. The participant was seated at a desk with the experimenter. The experimenter presented a variety of challenging academic task and provided verbal instructions to the participant. Contingent upon the occurrence of any target behaviour, the experimenter terminated the trial, turned away, and did not interact with the participant for 10 seconds following the cessation of the challenging behaviour. While praise for compliance and correct responding was delivered as usual throughout the condition, all other behaviours, excepting the target behaviours, were ignored. This condition was employed to determine whether any of the targeted challenging behaviours were sensitive to negative reinforcement in the form of escape from academic demand.

Tangible condition. The participant was provided with 30 seconds of access to preferred tangibles prior to commencement of this condition. At the beginning of the condition, the experimenter removed the tangibles from the participant and placed them within view of the participant. Ten seconds of access to these tangibles was provided contingent on the occurrence of a target behaviour. Following this, the experimenter took possession of the tangibles again. This condition was intended to assess whether the target behaviours were sensitive to positive reinforcement in the form of access to highly preferred stimuli.

Alone condition. The participant was placed in a room without access to social interaction or materials. The experimenter remained outside of the room and there were no programmed consequences for the occurrence of target behaviours. This condition was intended to assess whether the behaviour occurred in the absence of social contingencies and whether it may produce its own reinforcement.

Free play condition. During this condition, the participant had access to a variety of medium preference stimuli. The experimenter delivered attention on a fixed-schedule of 10 seconds. There were no demands placed upon the participant at any occasion. There were no programmed consequences for the occurrence of any target behaviours. This condition served as a control condition.

Transition condition (Participant 1 only). During this condition, the experimenter provided the participant with the instruction to transition to another room in the school. Following this, the experimenter delivered a similar instruction every 10 seconds (e.g., "Come along Participant 1, we are going to the playground!"). There were no programmed consequences for challenging behaviour during this condition. This condition was implemented to assess whether the demand of transitioning was associated with the rate of challenging behaviour for this participant.

Data Analysis

The functional analysis data was analysed by graphing the percentage of 10 second intervals during which the target challenging behaviour occurred. Subsequently, this data was analysed as per Martin, Gaffan and Williams (1997) guidelines for the interpretation of functional analysis data (see also Machalicek et al., 2014). Adapted from Hagopian et al.'s (1997) criteria for determining behavioural function from functional analysis outcomes, these guidelines for visual analysis involve the determination of the mean level of the target behaviour during the control condition (i.e., free play) and the plotting of a criterion line one standard deviation above the control condition mean. When behavioural frequency was at near-zero levels in the free play condition, the criterion line was set at 8.3% of intervals (Langthorne et al., 2011). Behavioural function was subsequently determined by identifying conditions in which at least one half of data points fell above the criterion line.

The cardiovascular and electrodermal data were analysed in a manner similar to that described in Study 3 (Lydon et al., 2015c/Chapter 4). However, differences in the experimental methodology and resulting data necessitated a number of changes to the previous data analysis methodology. First, participants' challenging behaviour occurred at such high frequencies during the five minute experimental conditions that it was not possible to extract a sufficient number of non-occurrences, defined in Study 3 (Lydon et al., 2015c/Chapter 4) as a 30 second period absent of challenging behaviour which was preceded and followed by a 30 second period during which no challenging behaviour was observed, in order to conduct a chi-square analysis to assess for differences in the HR or EDA patterns that co-occurred with challenging behaviour and non-occurrences of challenging behaviour. To compensate for this limitation, all occurrences of challenging behaviour, for which physiological data were available, were analysed rather than sampling occurrences as was carried out in Study 3 (Lydon et al., 2015c/Chapter 4).

The next difference in data analysis methodology related to the grouping of the occurrences of challenging behaviour for analysis. In Study 3 (Lydon et al., 2015c/Chapter 4), occurrences of challenging behaviour were grouped by participant mood state in order to assess for consistently co-occurring physiological waveforms as reported by Willemsen-Swinkels et al. (1998). However, given the null findings relating to mood state in the previous study, occurrences of challenging behaviour were not categorised by mood state in the current study. Instead, occurrences of challenging behaviour were categorised by the functional analysis condition during which they occurred (i.e., attention, escape, tangible, alone, free play, transitioning). This categorisation allowed for the assessment of whether behavioural function impacted on any physiological waveforms that co-occurred with challenging behaviour. Following this analysis, physiological waveforms that co-occurred with the target behaviours were assessed across all experimental conditions together. For both EDA and HR data, physiological patterns occurring as an antecedent to the behaviour

(i.e., 10 seconds physiological data prior to the onset of the behaviour as compared to the 10 seconds physiological data post-onset), as a consequent to the behaviour (i.e., 10 seconds physiological data prior to the offset of the behaviour as compared to 10 seconds physiological data post-offset), and the shift in HR or EDA as a result of engagement in the behaviour (i.e., 10 seconds physiological data prior to the onset of the behaviour as compared to 10 seconds physiological data post-offset of the behaviour) were assessed. Once again, changes in physiological activity were interpreted with regard to the SD of baseline physiological activity in order to detect any changes in physiological activity that was beyond that which would be expected. Changes in HR or EDA were therefore categorised as no change (i.e., change in HR or EDA that is less than or equivalent to the SD of that measure during the baseline period), increase (i.e., increase in HR or EDA is greater than the value of the SD of that measure during the baseline period), or decrease (i.e., decrease in HR or EDA that is greater than the value of the SD of that measure during the baseline period). HR and EDA patterns at each assessment timepoint (i.e., antecedent, consequent, and overall shift) were subsequently analysed. For the purposes of this research study, we considered HR or EDA patterns that co-occurred, at any of the three assessment timepoints, with more than 66.7% (two-thirds) of the occurrences of challenging behaviour to be meaningfully related. For the purposes of this research study, two-thirds of occurrences were considered to constitute a parsimonious preponderance of occurrences.

Following the analysis of physiological patterns that co-occurred with challenging behaviour, mean HR and mean EDA throughout incidents of challenging behaviour occurring during composure, elation, and distress mood states were assessed. As in Study 3 (Lydon et al., 2015c/Chapter 4), mean HR or EDA was calculated for each 12 second epoch within each occurrence of stereotypy. As all data arose from a single functional analysis conducted on one day, rather than across five days as in Study 3 (Lydon et al.,

2015c/Chapter 4), these data were investigated for each individual participant solely via Analysis of Variance. As in Study 3 (Lydon et al., 2015c/Chapter 4), each data-point was treated as a separate case for the purpose of this analysis. As described previously, ANOVA is based on an assumption of independent measures across multiple participants, an assumption violated in the current analysis, and the *p*-values resulting from the ANOVAs conducted in this study should be viewed as analogous to actual statistical significance levels. For each participant, two one-way between subjects ANOVAs with either HR or EDA as the dependent variable were conducted. Mood state with three levels (i.e., composure, elation, distress) served as the independent variable in these analyses.

Results

Participant 1

In total, 251 occurrences of challenging behaviour were recorded for Participant 1 across the six functional analysis conditions. Challenging behaviour was present during a total duration of 1 hour 16 minutes and 31 seconds (63.8% of total assessment). The mean duration of occurrences of challenging behaviour was 18 seconds (SD= 27 seconds; range 0-198 seconds). Occurrences were most frequently rated as mild (*n*=147; 58.6%), followed by moderate (*n*=104; 41.4%). Of the occurrences of challenging behaviour recorded, 166 occurred during composure (66.1%; 73.5% of these were rated as mild, and 26.5% were rated as moderate), 24 during elation (9.6%; 83.3% of occurrences were rated as mild, and 16.7% were rated as moderate), and 61 during periods of distress (24.3%; 9.8% of occurrences were mild, and 90.2% of occurrences were moderate). The results of Participant 1's functional analysis are presented in Figure 4. The application of Martin et al.'s (1997) guidelines for the interpretation of functional analysis results determined that SIB served a tangible function while fixing behaviours appeared to be maintained by negative reinforcement in the form of escape from demand and instructions to transition.

High levels of stereotyped motor behaviour across all conditions prevented the determination of behavioural function for this class of behaviours.

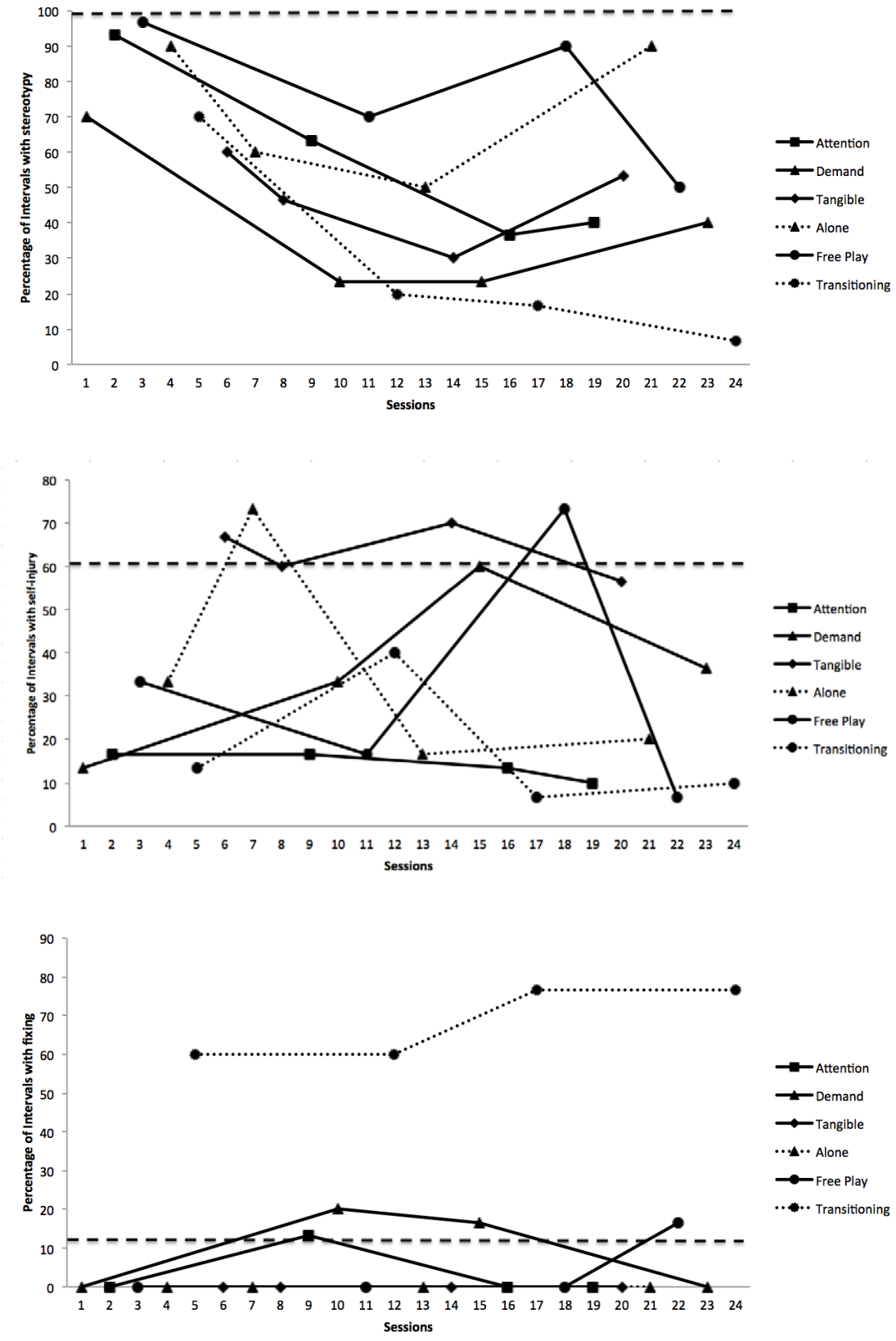

Figure 4. Functional analysis results for Participant 1's repetitive stereotyped behaviour (upper panel), self-injurious behaviour (middle panel), and fixing behaviours (lower panel).

Participant 1's mean HR was found to be 103.2 bpm (SD=16.1; range 49-178 bpm). HR patterns surrounding challenging behaviours were examined first. The outcomes of this analysis are presented in Table 16. None of the HR patterns met our criteria (>66.7%) for consistent occurrence.

Table 16

Heart Rate Patterns associated with Participant 1's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Challenging Behaviour (*n*=21)**	**Attention Condition**	**Occurrences of Challenging Behaviour (*n*=36)**	**Escape Condition**	**Occurrences of Challenging Behaviour (*n*=34)**	**Free Access (Play) Condition**	**Occurrences of Challenging Behaviour (*n*=29)**
Onset		**Onset**		**Onset**		**Onset**	
No change	*n*=13 (61.9%)	No change	*n*=16 (44.4%)	No change	*n*=13 (38.2%)	No change	*n*=10 (34.5%)
Increase	*n*=4 (19%)	Increase	*n*=13 (36.1%)	Increase	*n*=17 (50%)	Increase	*n*=12 (41.4%)
Decrease	*n*=4 (19%)	Decrease	*n*=7 (19.4%)	Decrease	*n*=4 (11.8%)	Decrease	*n*=7 (24.1%)
Offset		**Offset**		**Offset**		**Offset**	
No change	*n*=11 (52.4%)	No change	*n*=16 (44.4%)	No change	*n*=13 (38.2%)	No change	*n*=10 (34.5%)
Increase	*n*=5 (23.8%)	Increase	*n*=7 (19.4%)	Increase	*n*=7 (20.6%)	Increase	*n*=9 (31.0%)
Decrease	*n*=5 (23.8%)	Decrease	*n*=13 (36.1%)	Decrease	*n*=14 (41.2%)	Decrease	*n*=10 (34.5%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=7 (33.3%)	No change	*n*=12 (33.3%)	No change	*n*=13 (38.2%)	No change	*n*=9 (31.0%)
Increase	*n*=6 (28.6%)	Increase	*n*=14 (38.9%)	Increase	*n*=13 (38.2%)	Increase	*n*=11 (37.9%)
Decrease	*n*=8 (38.1%)	Decrease	*n*=10 (27.8%)	Decrease	*n*=8 (23.5%)	Decrease	*n*=9 (31.0%)

Tangible Condition	**Occurrences of Challenging Behaviour (*n*=34)**	**Transition Condition**	**Occurrences of Challenging Behaviour (*n*=8)**	**All Conditions**	**Occurrences of Challenging Behaviour (*n*=162)**
Onset		**Onset**		**Onset**	
No change	*n*=10 (29.4%)	No change	*n*=4 (50%)	No change	*n*=66 (40.1%)
Increase	*n*=17 (50%)	Increase	*n*=1 (12.5%)	Increase	*n*=64 (39.5%)
Decrease	*n*=7 (20.6%)	Decrease	*n*=3 (37.5%)	Decrease	*n*=32 (19.8%)
Offset		**Offset**		**Offset**	
No change	*n*=15 (44.1%)	No change	*n*=2 (25%)	No change	*n*=67 (41.4)
Increase	*n*=6 (17.6%)	Increase	*n*=5 (62.5%)	Increase	*n*=39 (24.1%)
Decrease	*n*=13 (38.2%)	Decrease	*n*=1 (12.5%)	Decrease	*n*=56 (34.6%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=5 (14.7%)	No change	*n*=1 (12.5%)	No change	*n*=47 (29%)
Increase	*n*=14 (41.2%)	Increase	*n*=4 (50%)	Increase	*n*=62 (38.3%)
Decrease	*n*=15 (44.1%)	Decrease	*n*=3 (37.5%)	Decrease	*n*=53 (32.7%)

Participant 1's mean EDA was found to be 0.67 µS (SD= 0.20; range 0.01-4.26 µS) across all sessions. The results of the examination of patterns of EDA surrounding occurrences of Participant 1's challenging behaviours are presented in Table 17. With the exception of "no change", none of the EDA patterns met our criteria for consistent occurrence at onset, offset, or in the overall change in physiological arousal associated with the challenging behaviour occurrence.

Table 17

Electrodermal Activity Patterns associated with Participant 1's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition Occurrences of Challenging Behaviour (*n*=35)		**Attention Condition** Occurrences of Challenging Behaviour (*n*=44)		**Escape Condition** Occurrences of Challenging Behaviour (*n*=32)		**Free Access (Play) Condition** Occurrences of Challenging Behaviour (*n*=35)	
Onset		**Onset**		**Onset**		**Onset**	
No change	*n*=19 (54.3%)	No change	*n*=27 (61.4%)	No change	*n*=23 (71.9%)	No change	*n*=26 (74.3%)
Increase	*n*=5 (14.3%)	Increase	*n*=6 (13.6%)	Increase	*n*= 1 (3.1%)	Increase	*n*= 3 (8.6%)
Decrease	*n*=11 (31.4%)	Decrease	*n*=11 (25%)	Decrease	*n*=8 (25%)	Decrease	*n*=6 (17.1%)
Offset		**Offset**		**Offset**		**Offset**	
No change	*n*=22 (62.9%)	No change	*n*=31 (70.5%)	No change	*n*=21 (65.6%)	No change	*n*=27 (77.1%)
Increase	*n*=4 (11.4%)	Increase	*n*=3 (6.8%)	Increase	*n*=1 (3.1%)	Increase	*n*=4 (11.4%)
Decrease	*n*=9 (25.7%)	Decrease	*n*=10 (22.7%)	Decrease	*n*=10 (31.3%)	Decrease	*n*=4 (11.4%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=18 (51.4%)	No change	*n*=23 (52.3%)	No change	*n*=17 (53.1%)	No change	*n*=22 (62.9%)
Increase	*n*=8 (22.9%)	Increase	*n*=4 (9.1%)	Increase	*n*=3 (9.4%)	Increase	*n*=7 (20%)
Decrease	*n*=9 (25.7%)	Decrease	*n*=17 (38.6%)	Decrease	*n*=12 (37.5%)	Decrease	*n*=6 (17.1%)

Tangible Condition Occurrences of Challenging Behaviour (*n*=40)		**Transition Condition** Occurrences of Challenging Behaviour (*n*=27)		**All Conditions** Occurrences of Challenging Behaviour (*n*=213)
Onset		**Onset**		**Onset**
No change	*n*=29 (72.5%)	No change	*n*=23 (85.2%)	No change	*n*=147 (69%%)
Increase	*n*= 4 (10%)	Increase	*n*= 2 (7.4%)	Increase	*n*=21 (9.9%)
Decrease	*n*=7 (17.5%)	Decrease	*n*=2 (7.4%)	Decrease	*n*=45 (21.1%)
Offset		**Offset**		**Offset**
No change	*n*=24 (60%)	No change	*n*=19 (70.4%)	No change	*n*=144 (67.6)
Increase	*n*=3 (7.5%)	Increase	*n*=5 (18.5%)	Increase	*n*=20 (9.4%)
Decrease	*n*=13 (32.5%)	Decrease	*n*=3 (11.1%)	Decrease	*n*=49 (23%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**
No change	*n*=23 (57.5%)	No change	*n*=12 (44.4%)	No change	*n*=115 (54%)
Increase	*n*=8 (20%)	Increase	*n*=12 (44.4%)	Increase	*n*=42 (19.7%)
Decrease	*n*=9 (22.5%)	Decrease	*n*=3 (11.1%)	Decrease	*n*=56 (26.3%)

Participant 1's level of HR and EDA during each of the three mood states (i.e., composure, elation, and distress) was also examined via one-way between subjects ANOVA. The analysis of HR during the different mood states revealed significant differences, $F(2,505)=16.43$, $p<.001$. HR during elation (M=89.5; SD=10.6) was significantly lower than HR during composure (M=100; SD=13.7) and distress (M=104.04; SD=12.1). A Tukey post-hoc test found that HR during distress was significantly higher than during composure. An ANOVA assessing for differences in EDA between the three mood states was also significant, $F(2,605)=38.0$, $p<.001$. As with HR, a Tukey post-hoc test revealed EDA was significantly lower during elation (M=.22; SD=.09) than during composure (M=.39; SD=.12) and distress (M=.43; SD=.16). EDA during composure was significantly lower than during distress.

Participant 2

In total, 109 occurrences of stereotyped motor behaviour were recorded for Participant 2 across the three functional analysis sessions conducted. The total duration of all occurrences of stereotyped motor behaviour was 37 minutes and 56 seconds (50.6% of total assessment duration). The mean duration of stereotypy was 21 seconds (SD= 28 seconds; range 1-148 seconds). Occurrences were most frequently rated as mild (n=92; 82.1%), followed by moderate (n=20; 17.9%). Of the occurrences recorded, 51 occurred during composure (46.8%; 96.1% of occurrences were mild, and 3.9% of occurrences were moderate), 49 during elation (45%; 91.8% were rated as mild, and 8.2% of occurrences were rated as moderate), and 9 during periods of distress (8.3%; all occurrences were rated as moderate). The results of Participant 2's functional analysis are presented in Figure 5. The application of Martin et al.'s (1997) guidelines for the interpretation of functional analysis failed to reveal a function of stereotyped motor behaviour.

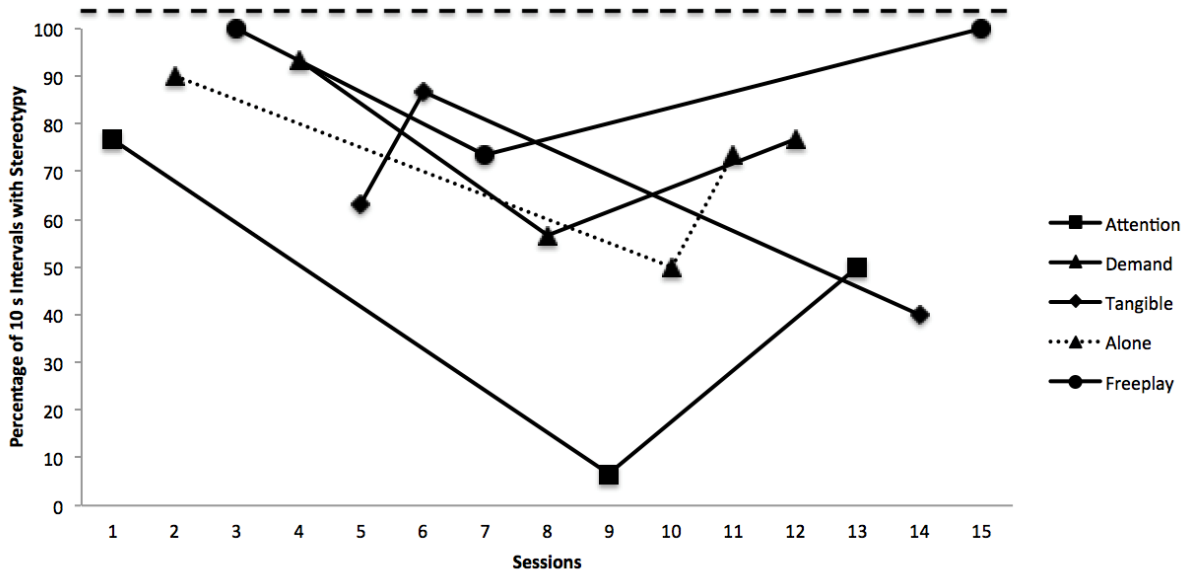

Figure 5. Functional analysis results for Participant 2's stereotyped behaviour.

Participant 2's mean HR was found to be 102.17 bpm (*SD*=22.29; range 61-200 bpm). HR patterns surrounding challenging behaviours were examined first. The outcomes of this analysis are presented in Table 18. None of the HR patterns met our criteria (>66.7%) for consistent occurrence.

Table 18

Heart Rate Patterns associated with Participant 2's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=19)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=5)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=20)**
Onset	*n*=9 (47.4%)	**Onset**	*n*=2 (40%)	**Onset**	
No change	*n*=6 (31.6%)	No change	*n*=2 (40%)	No change	*n*=6 (30%)
Increase	*n*=4 (21.1%)	Increase	*n*=1 (20%)	Increase	*n*=7 (35%)
Decrease		Decrease		Decrease	*n*=7 (35%)
Offset		**Offset**		**Offset**	
No change	*n*=6 (31.6%)	No change	*n*=1 (20%)	No change	*n*=9 (45%)
Increase	*n*=7 (36.8%)	Increase	*n*=1 (20%)	Increase	*n*=8 (40%)
Decrease	*n*=6 (31.6%)	Decrease	*n*=3 (60%)	Decrease	*n*=3 (15%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=4 (21.1%)	No change	*n*=2 (40%)	No change	*n*=2 (10%)
Increase	*n*=6 (31.6%)	Increase	*n*=0	Increase	*n*=11 (55%)
Decrease	*n*=9 (47.4%)	Decrease	*n*=3 (60%)	Decrease	*n*=7 (35%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=11)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=19)**	**All Conditions**	**Occurrences of Stereotypy (*n*=74)**
Onset		**Onset**		**Onset**	
No change	*n*=2 (18.2%)	No change	*n*=5 (26.3%)	No change	*n*=24 (32.4%)
Increase	*n*=6 (54.5%)	Increase	*n*=6 (31.6%)	Increase	*n*=27 (36.5%)
Decrease	*n*=3 (27.3%)	Decrease	*n*=8 (42.1%)	Decrease	*n*=23 (31.1%)
Offset		**Offset**		**Offset**	
No change	*n*=6 (54.5%)	No change	*n*=6 (31.6%)	No change	*n*=28 (37.8%)
Increase	*n*=3 (27.3%)	Increase	*n*=5 (26.3%)	Increase	*n*=24 (32.4%)
Decrease	*n*=2 (18.2%)	Decrease	*n*=8 (42.1%)	Decrease	*n*=22 (29.7%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=3 (27.3%)	No change	*n*=4 (21.2%)	No change	*n*=15 (20.3%)
Increase	*n*=6 (54.5%)	Increase	*n*=7 (36.8%)	Increase	*n*=30 (40.5%)
Decrease	*n*=2 (18.2%)	Decrease	*n*=8 (42.1%)	Decrease	*n*=29 (39.2%)

Participant 2's mean EDA was found to be 2.25 µS (SD= 0.66; range 0.06-6.26 µS) across all sessions. The results of the examination of patterns of EDA surrounding occurrences of Participant 2's stereotyped behaviours are presented in Table 19. Our analysis failed to reveal any pattern of EDA that was consistently associated (>66.7%) with the onset or offset of stereotypy, or an overall shift in EDA that consistently co-occurred with stereotypy.

Table 19

Electrodermal Activity Patterns associated with Participant 2's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=19)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=24)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=33)**
Onset		**Onset**		**Onset**	
No change	*n*=3 (15.8%)	No change	*n*=10 (41.7%)	No change	*n*=16 (48.5%)
Increase	*n*=7 (36.8%)	Increase	*n*=5 (20.8%)	Increase	*n*= 9 (27.3%)
Decrease	*n*=9 (47.4%)	Decrease	*n*=9 (37.5%)	Decrease	*n*=8 (24.2%)
Offset		**Offset**		**Offset**	
No change	*n*=6 (31.6%)	No change	*n*=12 (50%)	No change	*n*=18 (54.5%)
Increase	*n*=5 (26.3%)	Increase	*n*=7 (29.2%)	Increase	*n*=7 (21.2%)
Decrease	*n*=8 (42.1%)	Decrease	*n*=5 (20.8%)	Decrease	*n*=8 (24.2%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=3 (15.8%)	No change	*n*=5 (20.8%)	No change	*n*=13 (39.4%)
Increase	*n*=7 (36.8%)	Increase	*n*=7 (29.2%)	Increase	*n*=13 (39.4%)
Decrease	*n*=9 (47.4%)	Decrease	*n*=12 (50%)	Decrease	*n*=7 (21.2%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=15)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=20)**	**All Conditions**	**Occurrences of Stereotypy (*n*=111)**
Onset		**Onset**		**Onset**	
No change	*n*=3 (20%)	No change	*n*=7 (35%)	No change	*n*=39 (35.1%)
Increase	*n*= 2 (13.3%)	Increase	*n*= 6 (10%)	Increase	*n*= 29 (26.1%)
Decrease	*n*=10 (66.7%)	Decrease	*n*=7 (35%)	Decrease	*n*=43 (38.7%)
Offset		**Offset**		**Offset**	
No change	*n*=5 (33.3%)	No change	*n*=7 (35%)	No change	*n*=48 (43.2%)
Increase	*n*=4 (26.7%)	Increase	*n*=11 (55%)	Increase	*n*=34 (30.6%)
Decrease	*n*=6 (40%)	Decrease	*n*=2 (10%)	Decrease	*n*=29 (26.1%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=3 (20%)	No change	*n*=5 (25%)	No change	*n*=29 (26.1%)
Increase	*n*=4 (26.7%)	Increase	*n*=5 (25%)	Increase	*n*=36 (32.4%)
Decrease	*n*=8 (53.3%)	Decrease	*n*=10 (50%)	Decrease	*n*=46 (41.4%)

Participant 2's level of HR and EDA during each of the three mood states (i.e., composure, elation, and distress) was also examined via one-way between subjects ANOVA. The analysis of HR during the different mood states revealed significant differences, $F(2,258)=18.97$, $p<.001$. A Tukey post-hoc test found that HR during composure (M=85.2; SD=10.4) was significantly lower than HR during elation (M=94.6; SD=12.8) and distress (M=95.8; SD=13.6), p's<.001. HR during elation and distress did not differ significantly. A subsequent one-way between subjects ANOVA assessing for differences in EDA between the three mood states was non-significant, $F(2,279)=.538$, $p=.59$, indicating that EDA was similar throughout engagement in stereotyped behaviours associated with composure, elation, and distress.

Participant 3

In total, 116 occurrences of motor stereotypy were recorded for Participant 3 across the five functional analysis conditions. The total duration of all of these occurrences was 16 minutes and 48 seconds (16.8% of total assessment duration). The mean duration of occurrences of stereotypy was 9 seconds (SD= 10 seconds; range 1-63 seconds). Of these, 94 occurred during composure (81%; 91.5% of these were rated as mild, 8.5% were rated as moderate), 18 during elation (15.5%; 72.2% were rated as mild, 27.8% were rated as moderate), and four during periods of distress (3.4%; 50% were rated as mild, 50% were rated as moderate). Occurrences were most frequently rated as mild (n=101; 87.1%), followed by moderate (n=15; 12.9%). The results of Participant 3's functional analysis are presented in Figure 6. The application of Martin et al.'s (1997) guidelines for the interpretation of functional analysis results determined that Participant 3's motor stereotypy was multiply controlled, maintained by sources of positive reinforcement (attention and tangible), negative reinforcement (demand), and automatic reinforcement (alone).

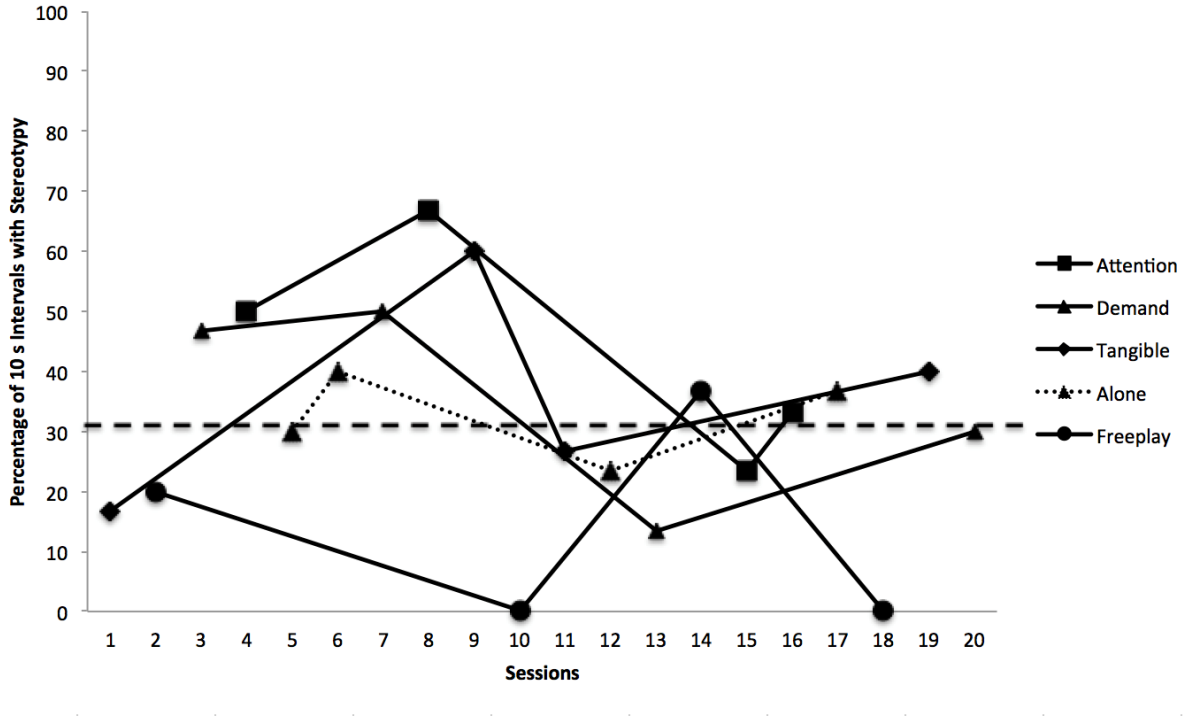

Figure 6. Functional analysis results for Participant 3's stereotyped behaviour.

Participant 3's mean HR was found to be 95.27 bpm (*SD*=15.3; range 61-165 bpm). HR patterns surrounding challenging behaviours were examined first. The outcomes of this analysis are presented in Table 20. None of the HR patterns met our criteria (>66.7%) for consistent occurrence.

Table 20

Heart Rate Patterns associated with Participant 3's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=21)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=30)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=26)**
Onset		**Onset**		**Onset**	
No change	*n*=8 (38.1%)	No change	*n*=10 (33.3%)	No change	*n*=7 (26.9%)
Increase	*n*=7 (33.3%)	Increase	*n*=9 (30%)	Increase	*n*=9 (34.6%)
Decrease	*n*=6 (28.6%)	Decrease	*n*=11 (36.7%)	Decrease	*n*=10 (38.5%)
Offset		**Offset**		**Offset**	
No change	*n*=5 (23.8%)	No change	*n*=9 (30%)	No change	*n*=5 (19.2%)
Increase	*n*=5 (23.8%)	Increase	*n*=5 (16.7%)	Increase	*n*=7 (26.9%)
Decrease	*n*=11 (52.4%)	Decrease	*n*=16 (53.3%)	Decrease	*n*=14 (53.8%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=4 (19%)	No change	*n*=9 (30%)	No change	*n*=6 (23.1%)
Increase	*n*=5 (23.8%)	Increase	*n*=12 (40%)	Increase	*n*=6 (23.1%)
Decrease	*n*=12 (57.1%)	Decrease	*n*=9 (30%)	Decrease	*n*=14 (53.8%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=11)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=26)**	**All Conditions**	**Occurrences of Stereotypy (*n*=114)**
Onset		**Onset**		**Onset**	
No change	*n*=4 (36.4%)	No change	*n*=8 (30.8%)	No change	*n*=37 (32.5%)
Increase	*n*=0	Increase	*n*=9 (34.6%)	Increase	*n*=34 (29.8%)
Decrease	*n*=7 (63.6%)	Decrease	*n*=9 (34.6%)	Decrease	*n*=43 (37.7%)
Offset		**Offset**		**Offset**	
No change	*n*=0	No change	*n*=10 (38.5%)	No change	*n*=29 (25.4%)
Increase	*n*=7 (63.6%)	Increase	*n*=9 (34.6%)	Increase	*n*=33 (28.9%)
Decrease	*n*=4 (36.4%)	Decrease	*n*=7 (26.9%)	Decrease	*n*=52 (45.6%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=4 (36.4%)	No change	*n*=16 (61.5%)	No change	*n*=39 (34.2%)
Increase	*n*=2 (18.2%)	Increase	*n*=5 (19.2%)	Increase	*n*=30 (26.3%)
Decrease	*n*=5 (45.5%)	Decrease	*n*=5 (19.2%)	Decrease	*n*=45 (39.5%)

Participant 3's mean EDA was found to be 0.84 µS (*SD*= 0.55; range 0.02-4.18 µS) across all sessions. The results of the examination of patterns of EDA surrounding occurrences of Participant 1's challenging behaviours are presented in Table 21. None of the EDA patterns surrounding occurrences of stereotypy met our criteria for consistent occurrence at onset, offset, or in the overall change in physiological arousal associated with the challenging behaviour occurrence.

Table 21

Electrodermal Activity Patterns associated with Participant 3's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=21)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=30)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=26)**
Onset		**Onset**		**Onset**	
No change	*n*=8 (38.1%)	No change	*n*=4 (13.3%)	No change	*n*=8 (30.8%)
Increase	*n*=3 (14.3%)	Increase	*n*=7 (23.3%)	Increase	*n*= 5 (19.2%)
Decrease	*n*=10 (47.6%)	Decrease	*n*=19 (63.3%)	Decrease	*n*=13 (50%)
Offset		**Offset**		**Offset**	
No change	*n*=8 (38.1%)	No change	*n*=5 (16.7%)	No change	*n*=10 (38.5%)
Increase	*n*=4 (19%)	Increase	*n*=10 (33.3%)	Increase	*n*=4 (15.4%)
Decrease	*n*=9 (42.9%)	Decrease	*n*=15 (50%)	Decrease	*n*=12 (46.2%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=7 (33.3%)	No change	*n*=11 (36.7%)	No change	*n*=7 (26.9%)
Increase	*n*=4 (19%)	Increase	*n*=9 (30%)	Increase	*n*=4 (15.4%)
Decrease	*n*=10 (47.6%)	Decrease	*n*=10 (33.3%)	Decrease	*n*=15 (57.7%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=11)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=26)**	**All Conditions**	**Occurrences of Stereotypy (*n*=114)**
Onset		**Onset**		**Onset**	
No change	*n*=5 (45.5%)	No change	*n*=5 (19.2%)	No change	*n*=30 (26.3%)
Increase	*n*= 2 (18.2%)	Increase	*n*= 4 (15.4%)	Increase	*n*= 21 (18.4%)
Decrease	*n*=4 (36.4%)	Decrease	*n*=17 (65.4%)	Decrease	*n*=63 (55.3%)
Offset		**Offset**		**Offset**	
No change	*n*=2 (18.2%)	No change	*n*=3 (11.5%)	No change	*n*=28 (24.6%)
Increase	*n*=3 (27.3%)	Increase	*n*=6 (23.1%)	Increase	*n*=27 (23.7%)
Decrease	*n*=6 (54.5%)	Decrease	*n*=17 (65.4%)	Decrease	*n*=59 (51.8%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=5 (45.5%)	No change	*n*=10 (38.5%)	No change	*n*=40 (35.1%)
Increase	*n*=4 (36.4%)	Increase	*n*=7 (26.9%)	Increase	*n*=28 (24.6%)
Decrease	*n*=2 (18.2%)	Decrease	*n*=9 (34.6%)	Decrease	*n*=46 (40.4%)

Participant 3's HR and EDA throughout stereotypy occurring during each of the three mood states (i.e., composure, elation, and distress) were also examined via one-way between subjects ANOVA. The analysis of HR during the different mood states revealed significant differences, $F(2,205)=55.13$, $p<.001$. A Tukey post-hoc test revealed that HR was significantly higher during elation ($M=106.2$; $SD=13.5$) than during composure ($M=89.8$; $SD=7.6$) or distress ($M=81.5$; $SD=3$), p's$<.001$. HR during composure and distress did not differ, $p=.25$. This pattern of findings was not replicated with Participant 3's electrodermal data; an ANOVA assessing for differences in EDA between the three mood states did not reveal significant differences between EDA during stereotypy occurring during the different mood states, $F(2,214)=1.67$, $p=.19$.

Participant 4

In total, 132 occurrences of challenging behaviour were recorded for Participant 4 across the five functional analysis conditions. The total duration of challenging behaviour was 55 minutes and 47 seconds (55.8% of total assessment duration). The mean duration of occurrences was 25 seconds (SD= 34 seconds; range 1-184 seconds). Of the occurrences of challenging behaviour recorded, 128 occurred during composure (97%; 78.9% were rated as mild, 21.1% were rated as moderate), and four occurred during elation (3%; 50% of these were rated as mild, 50% were rated as moderate). No occurrences of challenging behaviour were associated with distress. Occurrences were most frequently rated as mild ($n=102$; 76.7%), followed by moderate ($n=31$; 23.3%). The results of Participant 4's functional analysis are presented in Figure 7. The application of Martin et al.'s (1997) guidelines for the interpretation of functional analysis results determined that motor stereotypy (see upper panel Figure 7) was multiply controlled and sensitive to sources of positive reinforcement (tangible and attention), negative reinforcement (demand), and automatic reinforcement. As can be seen in the lower panel of Figure 7, repetitive SIB was found to be sensitive to negative reinforcement (escape).

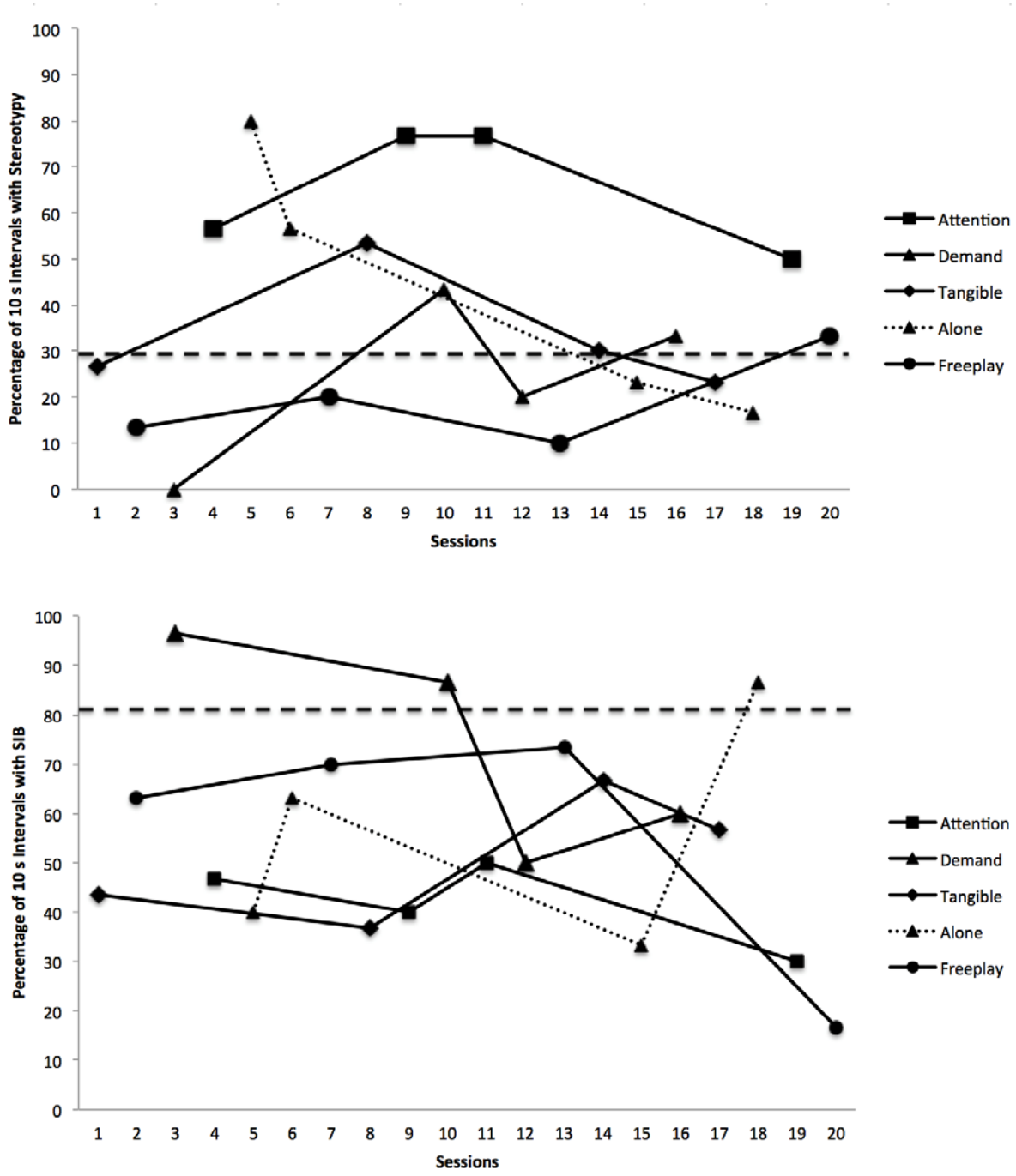

Figure 7. Functional analysis results for Participant 4's stereotyped behaviour (upper panel), and repetitive self-injurious behaviours (lower panel).

Participant 4's mean HR was found to be 100.88 bpm (*SD*=20.0; range 62-224 bpm). HR patterns surrounding challenging behaviours were examined first. The outcomes of this analysis are presented in Table 22. None of the HR patterns met our criteria (>66.7%) for

consistent occurrence. Mean HR during the various mood states was not examined for Participant 4 as almost all occurrences took place during composure (>96%) and the incidents occurring during elation (*n*=5; 3.8%) were considered to be insufficiently frequent to conduct the analysis.

Table 22

Heart Rate Patterns associated with Participant 4's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=12)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=30)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=23)**
Onset		**Onset**		**Onset**	
No change	*n*=3 (25%)	No change	*n*=10 (33.3%)	No change	*n*=7 (30.4%)
Increase	*n*=6 (50%)	Increase	*n*=12 (40%)	Increase	*n*=10 (43.5%)
Decrease	*n*=3 (25%)	Decrease	*n*=8 (26.7%)	Decrease	*n*=6 (26.1%)
Offset		**Offset**		**Offset**	
No change	*n*=6 (50%)	No change	*n*=16 (53.3%)	No change	*n*=7 (30.4%)
Increase	*n*=5 (41.7%)	Increase	*n*=6 (35.9%)	Increase	*n*=9 (39.1%)
Decrease	*n*=1 (8.3%)	Decrease	*n*=8 (15%)	Decrease	*n*=7 (30.4%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=4 (33.3%)	No change	*n*=5 (16.7%)	No change	*n*=7 (30.4%)
Increase	*n*=6 (50%)	Increase	*n*=14 (47.6%)	Increase	*n*=8 (34.8%)
Decrease	*n*=2 (16.7%)	Decrease	*n*=11 (36.7%)	Decrease	*n*=8 (34.8%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=15)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=45)**	**All Conditions**	**Occurrences of Stereotypy (*n*=125)**
Onset		**Onset**		**Onset**	
No change	*n*=7 (46.7%)	No change	*n*=6 (13.3%)	No change	*n*=33 (26.4%)
Increase	*n*=4 (26.7%)	Increase	*n*=27 (60%)	Increase	*n*=59 (47.3%)
Decrease	*n*=4 (26.7%)	Decrease	*n*=12 (26.7%)	Decrease	*n*=33 (26.4%)
Offset		**Offset**		**Offset**	
No change	*n*=4 (26.7%)	No change	*n*=17 (37.8%)	No change	*n*=50 (40%)
Increase	*n*=6 (40%)	Increase	*n*=8 (17.8%)	Increase	*n*=34 (27.2%)
Decrease	*n*=5 (33.3%)	Decrease	*n*=20 (44.4%)	Decrease	*n*=41 (32.8%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=4 (26.7%)	No change	*n*=15 (33.3%)	No change	*n*=35 (28%)
Increase	*n*=7 (46.7%)	Increase	*n*=23 (51.1%)	Increase	*n*=58 (46.4%)
Decrease	*n*=4 (26.7%)	Decrease	*n*=7 (15.6%)	Decrease	*n*=32 (25.6%)

Participant 5

In total, 168 occurrences of motor stereotypy were recorded for Participant 5 across the five functional analysis conditions. The total duration of all of these occurrences was 58 minutes and 41 seconds (58.7% of total assessment duration). The mean duration of occurrences of stereotypy was 21 seconds (*SD*= 27 seconds; range 1-161 seconds). Occurrences were most frequently rated as mild (*n*=147; 87.5%), followed by moderate (*n*=21; 12.5%). Of these, 112 occurred during composure (66.7%; 92.9% rated as mild, 7.1% rated as moderate), 51 during elation (30.4%; 82.5% rated as mild, 5.4% rated as moderate), and five during periods of distress (2.9%; 60% rated as mild, 40% rated as moderate). The results of Participant 5's functional analysis are presented in Figure 8. The application of Martin et al.'s (1997) guidelines for the interpretation of functional analysis results determined that Participant 5's motor stereotypy was multiply controlled, maintained by sources of positive reinforcement (attention and tangible), negative reinforcement (demand), and automatic reinforcement (alone).

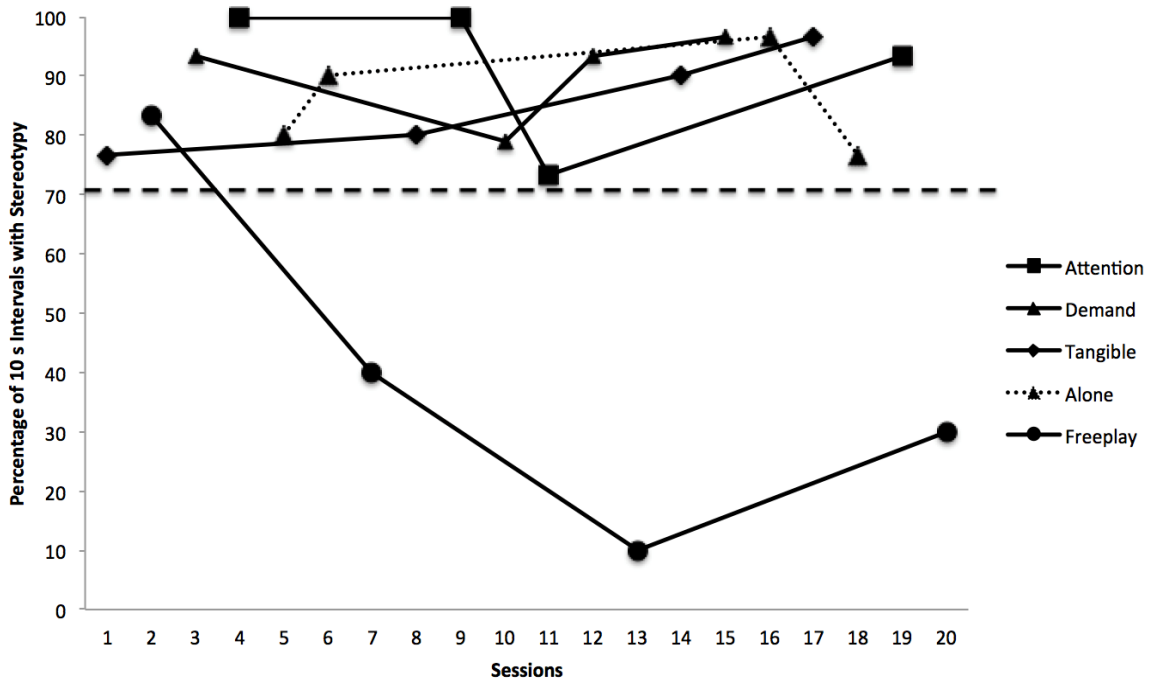

Figure 8. Functional analysis results for Participant 5's stereotyped behaviour.

Participant 5's mean HR was found to be 103.2 bpm (*SD*=11.5; range 71-173 bpm). HR patterns surrounding challenging behaviours were examined first. The outcomes of this analysis are presented in Table 23. None of the HR patterns met our criteria (>66.7%) for consistent occurrence.

Table 23

Heart Rate Patterns associated with Participant 5's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition		**Attention Condition**		**Escape Condition**	
	Occurrences of Stereotypy (*n*=17)		**Occurrences of Stereotypy (*n*=27)**		**Occurrences of Stereotypy (*n*=34)**
Onset		**Onset**		**Onset**	
No change	*n*=5 (29.4%)	No change	*n*=10 (37%)	No change	*n*=13 (38.2%)
Increase	*n*=3 (17.6%)	Increase	*n*=11 (40.7%)	Increase	*n*=16 (47.1%)
Decrease	*n*=9 (52.9%)	Decrease	*n*=6 (22.2%)	Decrease	*n*=5 (14.7%)
Offset		**Offset**		**Offset**	
No change	*n*=8 (47.1%)	No change	*n*=13 (48.1%)	No change	*n*=12 (35.3%)
Increase	*n*=4 (23.5%)	Increase	*n*=7 (25.9%)	Increase	*n*=9 (26.5%)
Decrease	*n*=5 (29.4%)	Decrease	*n*=7 (25.9%)	Decrease	*n*=13 (38.2%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=7 (41.2%)	No change	*n*=5 (18.5%)	No change	*n*=14 (41.2%)
Increase	*n*=3 (17.5%)	Increase	*n*=13 (48.1%)	Increase	*n*=14 (41.2%)
Decrease	*n*=7 (41.2%)	Decrease	*n*=9 (33.3%)	Decrease	*n*=6 (17.6%)
Free Access (Play) Condition		**Tangible Condition**		**All Conditions**	
	Occurrences of Stereotypy (*n*=20)		**Occurrences of Stereotypy (*n*=42)**		**Occurrences of Stereotypy (*n*=140)**
Onset		**Onset**		**Onset**	
No change	*n*=10 (50%)	No change	*n*=9 (21.4%)	No change	*n*=47 (33.6%)
Increase	*n*=6 (30%)	Increase	*n*=17 (40.5%)	Increase	*n*=53 (37.9%)
Decrease	*n*=4 (20%)	Decrease	*n*=16 (38.1%)	Decrease	*n*=40 (28.6%)
Offset		**Offset**		**Offset**	
No change	*n*=10 (50%)	No change	*n*=15 (35.7%)	No change	*n*=58 (41.4%)
Increase	*n*=3 (15%)	Increase	*n*=15 (35.7%)	Increase	*n*=38 (27.1%)
Decrease	*n*=7 (35%)	Decrease	*n*=12 (28.6%)	Decrease	*n*=44 (31.4%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=6 (30%)	No change	*n*=18 (42.9%)	No change	*n*=50 (35.7%)
Increase	*n*=6 (30%)	Increase	*n*=16 (38.1%)	Increase	*n*=52 (37.1%)
Decrease	*n*=8 (40%)	Decrease	*n*=8 (19%)	Decrease	*n*=38 (27.1%)

Participant 5's mean EDA was found to be 0.14 µS (*SD*= 0.09 range 0.01-1.17 µS) across all sessions. The results of the examination of patterns of EDA surrounding occurrences of Participant 5's challenging behaviours are presented in Table 24. None of the EDA patterns surrounding occurrences of stereotypy met our criteria for consistent occurrence at onset, offset, or in the overall change in physiological arousal associated with the challenging behaviour occurrence.

Table 24

Electrodermal Activity Patterns associated with Participant 5's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=14)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=27)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=33)**
Onset		**Onset**		**Onset**	
No change	*n*=6 (42.9%)	No change	*n*=11 (40.7%)	No change	*n*=16 (48.5%)
Increase	*n*=7 (50%)	Increase	*n*=5 (18.5%)	Increase	*n*= 14 (42.4%)
Decrease	*n*=1 (7.1%)	Decrease	*n*=11 (40.7%)	Decrease	*n*=3 (9.1%)
Offset		**Offset**		**Offset**	
No change	*n*=6 (42.9%)	No change	*n*=9 (33.3%)	No change	*n*=21 (63.6%)
Increase	*n*=5 (35.7%)	Increase	*n*=11 (40.7%)	Increase	*n*=2 (6.1%)
Decrease	*n*=3 (21.4%)	Decrease	*n*=7 (25.9%)	Decrease	*n*=10 (30.3%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=3 (21.4%)	No change	*n*=7 (25.9%)	No change	*n*=9 (27.3%)
Increase	*n*=8 (57.1%)	Increase	*n*=12 (44.4%)	Increase	*n*=19 (57.6%)
Decrease	*n*=3 (21.4%)	Decrease	*n*=8 (29.6%)	Decrease	*n*=5 (15.2%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=20)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=42)**	**All Conditions**	**Occurrences of Stereotypy (*n*=136)**
Onset		**Onset**		**Onset**	
No change	*n*=9 (45%)	No change	*n*=18 (42.9%)	No change	*n*=60 (44.1%)
Increase	*n*= 6 (30%)	Increase	*n*= 16 (38.1%)	Increase	*n*= 48 (35.3%)
Decrease	*n*=5 (25%)	Decrease	*n*=8 (19%)	Decrease	*n*=28 (20.6%)
Offset		**Offset**		**Offset**	
No change	*n*=11 (55%)	No change	*n*=19 (45.2%)	No change	*n*=66 (48.5%)
Increase	*n*=5 (25%)	Increase	*n*=19 (45.2%)	Increase	*n*=42 (30.9%)
Decrease	*n*=4 (20%)	Decrease	*n*=4 (9.5%)	Decrease	*n*=28 (20.6%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=6 (30%)	No change	*n*=12 (28.6%)	No change	*n*=37 (27.2%)
Increase	*n*=8 (40%)	Increase	*n*=24 (57.1%)	Increase	*n*=71 (52.2%)
Decrease	*n*=6 (30%)	Decrease	*n*=6 (14.3%)	Decrease	*n*=28 (20.6%)

Participant 5's HR and EDA throughout stereotypy occurring during each of the three mood states (i.e., composure, elation, and distress) were also examined via one-way between subjects ANOVA. The analysis of HR during the different mood states revealed no significant differences, $F(2,445)=.92$, $p=.40$, although HR during distress (M=99.6; SD=7.4) was lower than HR during composure (M=101.6; SD=5.9) and elation (M=101.8; SD=7.1). This pattern of findings was replicated with Participant 5's electrodermal data; an ANOVA assessing for differences in EDA between the three mood states revealed significant differences between EDA during stereotypy occurring during the different mood states, $F(2,361)=18.23$, $p<.001$. A Tukey post-hoc test revealed that EDA during elation (M=.19; SD=.1) was significantly higher than EDA during composure (M=.16; SD=.08), $p=.001$, or during distress (M=.06; SD= .02), $p<.001$. Further, HR during composure was found to be significantly higher than HR during distress, $p<.001$.

Participant 6

In total, 86 occurrences of motor stereotypy were recorded for Participant 6 across the five functional analysis conditions. The total duration of all of these occurrences was 1 hour, 38 minutes and 53 seconds (98.9% of total assessment duration). The mean duration of occurrences of stereotypy was 62 seconds (SD= 1 minute and 3 seconds; range 0-300 seconds). Of these, 50 occurred during elation (58.1%; 18% were rated as mild, 64% were rated as moderate, and 18% were rated as severe) and 36 occurred during composure (41.9%; 33% were rated as mild, 55.6% were rated as moderate, 11.1% were rated as severe). There were no occurrences of stereotypy associated with distress. Occurrences were most frequently rated as moderate (n=51; 59.3%), followed by mild (n=21; 24.4%), and severe (n=14; 16.3%). The results of Participant 6's functional analysis are presented in Figure 9. The results of the functional analysis were undifferentiated and it was not possible to identify the function of Participant 6's motor stereotypy.

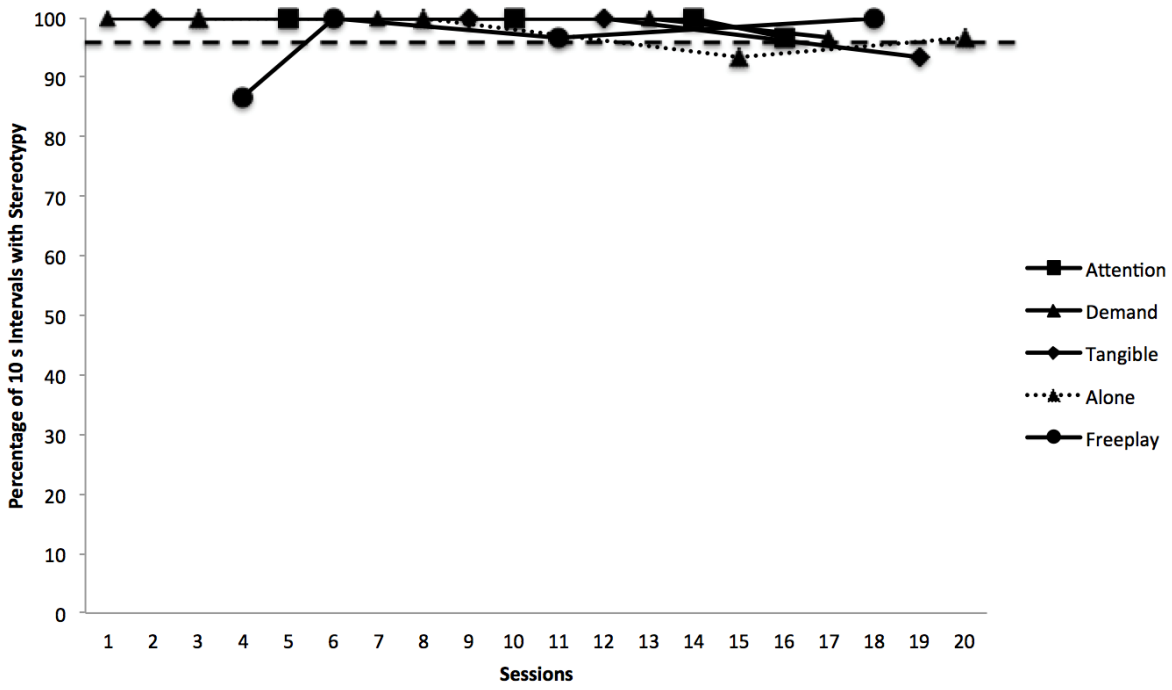

Figure 9. Functional analysis results for Participant 6's stereotyped behaviour.

Participant 6's mean HR was found to be 105.7 bpm (*SD*=11.7; range 68-173 bpm). HR patterns surrounding challenging behaviours were examined first. The outcomes of this analysis are presented in Table 25. None of the HR patterns met our criteria (>66.7%) for consistent occurrence. Mean HR during stereotypy associated with each of the three mood states was not assessed via one-way between subjects ANOVA for participant as stereotypy occurred only during elation and composure with no occurrences of stereotypy recorded during distress.

Table 25

Heart Rate Patterns associated with Participant 6's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition		**Attention Condition**		**Escape Condition**	
	Occurrences of Stereotypy (*n*=9)		**Occurrences of Stereotypy (*n*=21)**		**Occurrences of Stereotypy (*n*=17)**
Onset		**Onset**		**Onset**	
No change	*n*=2 (22.2%)	No change	*n*=5 (23.8%)	No change	*n*=9 (52.9%)
Increase	*n*=5 (55.6%)	Increase	*n*=7 (33.3%)	Increase	*n*=3 (17.6%)
Decrease	*n*=2 (22.2%)	Decrease	*n*=9 (42.9%)	Decrease	*n*=5 (29.4%)
Offset		**Offset**		**Offset**	
No change	*n*=3 (33.3%)	No change	*n*=9 (42.9%)	No change	*n*=11 (64.7%)
Increase	*n*=4 (44.4%)	Increase	*n*=5 (23.8%)	Increase	*n*=3 (17.6%)
Decrease	*n*=2 (22.2%)	Decrease	*n*=7 (33.3%)	Decrease	*n*=3 (17.6%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=4 (44.4%)	No change	*n*=4 (19%)	No change	*n*=5 (29.4%)
Increase	*n*=2 (22.2%)	Increase	*n*=8 (38.1%)	Increase	*n*=5 (29.4%)
Decrease	*n*=3 (33.3%)	Decrease	*n*=9 (42.9%)	Decrease	*n*=7 (41.2%)
Free Access (Play) Condition		**Tangible Condition**		**All Conditions**	
	Occurrences of Stereotypy (*n*=17)		**Occurrences of Stereotypy (*n*=17)**		**Occurrences of Stereotypy (*n*=81)**
Onset		**Onset**		**Onset**	
No change	*n*=8 (47.1%)	No change	*n*=5 (29.4%)	No change	*n*=29 (35.8%)
Increase	*n*=4 (23.5%)	Increase	*n*=8 (47.1%)	Increase	*n*=27 (58.4%)
Decrease	*n*=5 (29.4%)	Decrease	*n*=4 (23.5%)	Decrease	*n*=25 (30.9%)
Offset		**Offset**		**Offset**	
No change	*n*=8 (47.1%)	No change	*n*=6 (35.3%)	No change	*n*=37 (45.7%)
Increase	*n*=5 (29.4%)	Increase	*n*=5 (29.4%)	Increase	*n*=22 (27.2%)
Decrease	*n*=4 (23.5%)	Decrease	*n*=6 (35.3%)	Decrease	*n*=22 (27.2%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=5 (29.4%)	No change	*n*=8 (47.1%)	No change	*n*=26 (32.1%)
Increase	*n*=5 (29.4%)	Increase	*n*=4 (23.5%)	Increase	*n*=24 (29.6%)
Decrease	*n*=7 (41.2%)	Decrease	*n*=5 (29.4%)	Decrease	*n*=31 (38.3%)

Participant 6's mean EDA was found to be 0.79 µS (*SD*= 0.55; range 0.006-7.22 µS) across all sessions. The results of the examination of patterns of EDA surrounding occurrences of Participant 1's challenging behaviours are presented in Table 26. None of the EDA patterns surrounding occurrences of stereotypy met our criteria for consistent occurrence. As with HR data, mean EDA was not assessed across mood states due to the lack of occurrences associated with distress.

Table 26

Electrodermal Activity Patterns associated with Participant 6's Repetitive, Stereotyped Behaviours across Functional Analysis Conditions

Alone Condition	**Occurrences of Stereotypy (*n*=13)**	**Attention Condition**	**Occurrences of Stereotypy (*n*=20)**	**Escape Condition**	**Occurrences of Stereotypy (*n*=17)**
Onset		**Onset**		**Onset**	
No change	*n*=5 (38.5%)	No change	*n*=9 (45%)	No change	*n*=5 (29.4%)
Increase	*n*=3 (23.1%)	Increase	*n*=1 (5%)	Increase	*n*= 2 (11.8%)
Decrease	*n*=5 (38.5%)	Decrease	*n*=10 (50%)	Decrease	*n*=10 (58.8%)
Offset		**Offset**		**Offset**	
No change	*n*=5 (38.5%)	No change	*n*=8 (40%)	No change	*n*=7 (41.2%)
Increase	*n*=4 (30.8%)	Increase	*n*=4 (20%)	Increase	*n*=2 (11.8%)
Decrease	*n*=4 (30.8%)	Decrease	*n*=8 (40%)	Decrease	*n*=8 (47.1%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=3 (23.1%)	No change	*n*=5 (25%)	No change	*n*=2 (11.8%)
Increase	*n*=3 (23.1%)	Increase	*n*=6 (30%)	Increase	*n*=5 (29.4%)
Decrease	*n*=7 (53.8%)	Decrease	*n*=9 (45%)	Decrease	*n*=10 (58.8%)
Free Access (Play) Condition	**Occurrences of Stereotypy (*n*=17)**	**Tangible Condition**	**Occurrences of Stereotypy (*n*=17)**	**All Conditions**	**Occurrences of Stereotypy (*n*=84)**
Onset		**Onset**		**Onset**	
No change	*n*=6 (35.3%)	No change	*n*=5 (29.4%)	No change	*n*=30 (35.7%)
Increase	*n*=4 (23.5%)	Increase	*n*= 3 (17.6%)	Increase	*n*=13 (15.5%)
Decrease	*n*=7 (41.2%)	Decrease	*n*=9 (52.9%)	Decrease	*n*=41 (48.8%)
Offset		**Offset**		**Offset**	
No change	*n*=5 (29.4%)	No change	*n*=5 (29.4%)	No change	*n*=30 (35.7%)
Increase	*n*=3 (17.6%)	Increase	*n*=3 (17.6%)	Increase	*n*=16 (19%)
Decrease	*n*=9 (52.9%)	Decrease	*n*=9 (52.9%)	Decrease	*n*=38 (45.2%)
Overall change in physiological arousal associated with stereotypy		**Overall change in physiological arousal associated with stereotypy**		**Overall change in physiological arousal associated with stereotypy**	
No change	*n*=1 (5.9%)	No change	*n*=2 (11.8%)	No change	*n*=13 (15.5%)
Increase	*n*=5 (29.4%)	Increase	*n*=7 (41.2%)	Increase	*n*=26 (31%)
Decrease	*n*=11 (64.7%)	Decrease	*n*=8 (47.1%)	Decrease	*n*=45 (53.6%)

Discussion

The current study sought to evaluate the utility of a model of experimental functional analysis incorporating measures of physiological activity for assessing problematic repetitive, stereotyped behaviours that were either automatically reinforced or which had been resistant to prior function-based behavioural intervention. An additional aim was to assess physiological activation during repetitive, stereotyped behaviour associated with various mood states in order to assess for the presence of physiological blunting, such as that observed in Study 3 (Lydon et al., 2015c/Chapter 4). Across the six children and adolescents who took part, there appeared to be no relation between HR or EDA and engagement in repetitive, stereotyped behaviour. Further, a level of physiological activation suggestive of blunting was observed for only two participants. These results contribute to the scholarly discussion surrounding the utility of functional analyses incorporating physiological measures and to our knowledge of physiological activity in ASD.

A number of researchers have called for the utilisation of physiological measures during the experimental functional analysis of challenging behaviour to be examined empirically (Baker et al., 2002; Cohen et al., 2011; Romanczyk & Matthews, 1998) given the growing body of research suggestive of some association between patterns, or levels, of physiological activity and engagement in challenging behaviour (for reviews, see: Chapter 1; Cohen et al., 2011). The current study sought to present a practitioner-friendly model for conducting functional analysis while capturing measures of HR and EDA and for analysing the resultant data. Participants were selected on the basis of their engagement in problematic repetitive, stereotyped behaviours that had been resistant to previous behavioural intervention or which were identified by indirect assessment (i.e., the QABF) to be automatically reinforced. These participants were considered to present with "difficult" challenging behaviours, for which typical behavioural assessments were

unlikely to aid with the development of effective function-based behavioural interventions. It was hoped that the addition of measures of physiological activity to the functional analysis of the challenging behaviour exhibited by these participants would provide a greater understanding of their behaviour and suggest treatment approaches that could not typically be recommended from the results of a functional analysis (e.g., antecedent exercise, relaxation strategies, functional communication training to request high arousal activities or low arousal activities as appropriate).

The results from our analyses of physiological activity surrounding participants' challenging behaviour are indicative of little association between EDA and HR and engagement in repetitive, stereotyped behaviours however. For these participants, the addition of physiological measures to the functional analysis of their challenging behaviour appeared to add no incremental validity to the standard functional analysis nor did it yield suggestions for treatment that would not otherwise have been considered. These findings are not wholly surprising in light of the findings of Study 3 (Lydon et al., 2015c/Chapter 4), however they are in contrast to the many theories (Barrera et al., 2007; Groden et al., 1994; Guess & Carr, 1991; Hutt et al., 1964; Lydon et al., 2013; Romanczyk, 1986; Sugarman et al., 2014; Tordjman et al., 2014), and the sizeable body of supporting research (for a review, see Cohen et al., 2011; Chapter 1), linking physiological activity and engagement in stereotyped and self-injurious challenging behaviour. However, in light of the inconsistencies in the findings of psychophysiological research studies conducted among persons with autism (Benevides & Lane, 2013; Klusek et al., 2014; Lydon et al., in press/Study 1: Chapter 2; Taylor & Corbett, 2014), the failure to replicate the findings of other psychophysiological research studies, including a study conducted by our own research team (Lydon et al., 2013), is perhaps not unexpected. These inconsistencies in research findings once again highlight the need for the further investigation of possible physiological profiles of individuals with ASD (i.e., subtypes of ASD presenting with

distinct differences in baseline levels of physiological activity and PR to stimuli). The identification of distinct physiological profiles of ASD, the existence of which has been proposed by previous researchers (Hirstein et al., 2001; Schoen et al., 2008; Lydon et al., in press/Study 1: Chapter 2), who present with different behavioural or cognitive profiles would be a significant contribution to the research literature with persons diagnosed with autism and may also aid with the elucidation of inconsistent research findings resulting from psychophysiological studies of persons with ASD. Further, characterising the functional abilities, health states, and behavioural manifestations of potential subtypes of individuals with ASD who present with different physiological profiles would be of great interest. Such an analysis would provide a wealth of information regarding potential moderators of PR (e.g., cognitive ability, verbal status, adaptive behaviour), and further information on the relation between PR and adaptive and maladaptive behaviour. Without homogenising samples of individuals with ASD in some manner for psychophysiological research studies, it is likely that conflicting findings will continue to result and will impede our development of a fuller understanding of the psychophysiology of autism.

The findings of the current study regarding the lack of association between physiological activity and engagement in repetitive behaviour must also prompt consideration and discussion of automatic reinforcement processes among behaviour analysts working with individuals diagnosed with autism. Automatic reinforcement has long been considered one of the more problematic concepts of behaviour theory (Barrera et al., 2007; Vaughan & Michael, 1982). Although useful assessment processes for identifying the source of automatic reinforcement have been described in the literature (Rincover et al., 1979; Tang et al., 2003), behavioural interventions for repetitive, stereotyped behaviour are typically less effective than behavioural interventions for other forms of challenging behaviour (Rapp et al., 2005a), and stereotyped behaviours are the type of challenging behaviour most commonly treated using punishment procedures in the

research literature from recent decades (Lydon et al., 2015a). These findings indicate a continuing struggle among behaviour analysts to reduce engagement in repetitive behaviours among persons with autism. In the current study, participants engaged in a variety of repetitive behaviours that caused physical harm to their own bodies, interfered with their ability to engage with academic instruction and to make progress towards their learning objectives, and resulted in disruption to their home environment. There was unanimous agreement amongst their services and families that a reduction in the rates of engagement in these behaviours would be beneficial for the individual's quality of life. However, previous behavioural and pharmacological interventions to reduce the rates of engagement in these behaviours had been unsuccessful. It is clear that further research focused on elucidating the various potential sources of stimulation (e.g., auditory, visual, tactile, proprioceptive, etc.) functioning as automatic reinforcement for such behaviours is necessary. Such research would offer a significant contribution to the field of behaviour analysis, particularly if accompanied by treatment evaluations and treatment recommendations for the various types of sensory stimulation that may play a role in the perpetuation of such behaviours. The dissemination of the methods and outcomes of such research among the behavioural community may improve behaviour analysts' ability to reduce repetitive behaviours, decrease the reliance on punishment-based procedures and improve the academic and social engagement of persons with autism over time.

Levels of physiological activity during repetitive, stereotyped behaviour associated with each of the three mood states (i.e., elation, composure, and distress) were examined for Participants 1, 2, 3, and 5. Participants 4 and 6 engaged in repetitive, stereotyped behaviour during one mood state predominantly and were excluded from these analyses as a result. The results of Study 3 (Lydon et al., 2015c/Chapter 4) were suggestive of physiological blunting among three of the four participants with ASD who were included in this analysis. Data arising from these participants indicated that HR during occurrences

of motor stereotypy associated with distress was similar to, or lower than, HR during occurrences of motor stereotypy associated with outward composure. This finding was somewhat unexpected as previous research has indicated that physiological activation typically increases during distress (Pressman & Cohen, 2005) and was suggestive of physiological blunting in response to stressors. However, Lydon et al. (2013) had previously noted low HR during engagement in challenging behaviour evoked by stressors. Given the negative outcomes associated with physiological blunting (al'Absi et al., 2005; Lovallo et al., 2000; Ortiz & Raine, 2004; Phillips et al., 2012), the current research study sought to further investigate physiological activation during distress among the participants included in the current study, in order to determine whether participants in this sample also exhibited patterns of electrodermal and HR activity during distress that were indicative of physiological blunting. Our results indicated that for Participants 1 and 2, HR was higher during distress than during composure, a typical stress response. However, HR during occurrences of stereotypy associated with composure and distress were similar, and lower than HR during elated occurrences, for Participants 3 and 5, although this finding was only significant for Participant 3, a pattern potentially suggestive of physiological blunting. EDA data showed less differentiation between the mood states, with similar levels of EDA across the three mood states observed for Participants 2 and 3. For Participant 1, however, EDA was higher during distressed occurrences than during occurrences associated with composure while for Participant 5 EDA during distress was significantly lower than EDA during elation or composure. These results, which do not appear to be explained by an uneven distribution of mild, moderate, or severe stereotypy during any of the mood states, suggest that physiological blunting may exist for a subset of individuals with ASD (i.e., Participants in Study 3 (Lydon et al., 2015c/Chapter 4) and Participants 3 and 5 in the current study). Such a finding provides a tentative suggestion of a sub-type of ASD which requires further investigation and empirical validation, but is unlikely to characterise the

PR patterns of all persons with ASD. This in line with the systematic review of PR to stimuli among persons with autism conducted in Study 1 (Lydon et al., in press/ Chapter 2) that identified inconsistent findings regarding PR to stressor stimuli amongst samples of individuals with ASD. Further, among typically developing persons physiological blunting characterises the response patterns of only a portion of persons (Lovallo, 2011). However, while not present for all individuals with ASD, the current research program does highlight that this pattern of PR may be relatively common among individuals with ASD (3/5 participants in Study 3 (Lydon et al., 2015c/Chapter 4), and 2/4 participants in the current study) and the prevalence and implications of this do warrant exploration in future research studies. Study 1 (Lydon et al., in press/Chapter 2) revealed that research examining PR to stressors among persons with autism has typically excluded lower functioning individuals, has been conducted primarily in laboratory settings rather than the natural environment, and has focused upon stressors designed for typically developing populations rather than individuals with autism. Future research must therefore endeavour to remedy these deficits by studying PR among lower functioning individuals with ASD, in their natural environments, and in response to stressors individualised for participants.

The current study had a number of strengths including the determination of behavioural function using an experimental functional analysis methodology allowing for the more confident assertion that the behaviours under assessment were at least partially maintained by automatic sources of reinforcement. The concurrent utilisation of two measures of physiological activity during the functional analysis procedures added strength to the study, given that previous research has relied solely upon one measure of physiological activity and has drawn criticism as a result (Cohen et al., 2011). The current study also adhered to best practice for the inclusion of children with autism in psychophysiological research studies (Kylliainen et al., 2014). However, there were also a number of limitations to the current study that must be considered when interpreting the

findings. First, for Participant 2 only three of the intended four repetitions of functional analysis conditions were conducted. This decision was made due to escalations in this participant's aggressive behaviours that endangered others in the experimental environment. However, the early termination of the functional analysis resulted in reduced data for this participant and it is possible that functional analysis outcomes or physiological data analysis outcomes may have differed had the functional analysis, and data collection, been completed as intended. Second, Participant 3 was identified by his parents and instructors to present with high levels of vigorous motor stereotypy. These high levels of engagement were verified by the primary researcher during the initial habituation phase for this participant. However, Participant 3 exhibited low levels of motor stereotypy across the functional analysis conditions (only 16.8% of total assessment duration was devoted to engagement in stereotyped behaviour), and the majority of motor stereotypy observed was comparatively mild. It is unclear why engagement in stereotypy decreased in frequency and severity during the experimental period; it is possible that Participant 3's stereotypy was maintained by a reinforcing contingency not assessed during the functional analysis. Alternatively, while the functional analysis was conducted in the participant's natural environment, it is possible that novel elements such as the presence of two members of the research team and the video recording of the sessions may have impacted on Participant 3's behaviour. The low levels of stereotyped behaviour, and the mild nature of such variables, negatively impacted on our analysis for this participant by reducing the number of occurrences available for analysis and calling into question the representativeness of the occurrences recorded as compared to his typical stereotyped behaviour. A third limitation was the substantial proportion of missing HR and EDA data noted during the analysis of the physiological data. This was due to the movement or disruption of the HR monitor by the participants or the malfunctioning of the EDA monitor during data collection. Kylliainen and colleagues (2014) have previously remarked upon the near impossibility of

conducting psychophysiological experiments with children with ASD without encountering the challenges related to portable equipment and the issue of missing data. Further, the authors of other similar research studies have also noted high frequencies of equipment malfunctioning and missing physiological data (Hoch et al., 2013; Moskowitz et al., 2013; Romanczyk & Matthews, 1998). Thus, the degree of missing physiological data observed in the current study is unfortunate but not an anomaly in the research literature.

Finally, the method of data analysis employed in the current study may also be critiqued. The five minute functional analysis condition durations employed coupled with the high rates and long durations of participants' repetitive, stereotyped behaviours precluded the extraction of non-occurrences of repetitive, stereotyped behaviours and the comparison of physiological patterns surrounding occurrences and non-occurrences via chi-square test as in Study 3 (Lydon et al., 2015c/Chapter 4). The possibility of extracting non-occurrences from physiological data collected outside of the experimental functional analysis was dismissed as it was considered that the functional analysis may have constituted a stressor for participants and resulted in physiological activation that differed from typical activation during participants' everyday routines resulting in an inappropriate sample of non-occurrences for analysis. The criteria for significance or noteworthiness (>66.7%) was considered stringent by the researchers and likely to identify only physiological patterns that were reliably and consistently associated with the onset or offset of occurrences, or resulting from occurrences. However, the arbitrary designation of 66.7% as the point at which an "out of the ordinary effect" would be recognised may also be criticised, or considered a limitation of the study. As emphasised in Study 3 (Lydon et al., 2015c/Chapter 4) however, the method of data analysis employed in the current study was considered both a sound means of addressing our research questions and also a methodology that would be accessible to, and feasible for, practitioners who may choose to

employ such psychophysiological assessments. Future research may wish to evaluate alternate methods of analysis for data such as that collected in the current study that may offer a more efficient or more effective means of analysing data collected in the context of psychophysiological assessments of challenging behaviour.

Chapter 6

General Discussion

Summary of Studies

The current research program aimed to address Romanczyk and Matthew's (1998) question regarding engagement in challenging behaviour by persons with autism and other developmental disabilities. Specificially, "what is the explanatory value of physiological data, and what new information can the data provide?" (p. 117). A systematic review and a series of empirical studies examined various indices of physiological arousal among children and adolescents diagnosed with ASD, and investigated the association between physiological activity and challenging behaviour for these individuals.

Study 1

The first study comprised a systematic review of five decades of extant research on PR to sensory, social and emotional, and stressor stimuli among persons with autism. The results revealed that individuals with autism differed from typically developing persons on measures of PR in 78.6% of studies employing sensory stimuli, 66.7% of studies using social or emotional stimuli, and 71.4% of studies utilising stressor stimuli. A number of reviewed studies were suggestive of physiological dysregulation of the LHPA axis and vagal systems in autism. However, the study found the existing body of research to be characterised by highly variable and inconsistent outcomes across the reviewed studies. Several of the studies reviewed were suggestive of the existence of physiological sub-groups among the autism group, including hypo-aroused and hyper-aroused individuals, the existence of which may impact upon the outcomes of studies of PR and physiological activity in autism.

Study 2

Study 2 (Lydon et al., 2015b/Chapter 3) assessed stress levels, through the measurement of diurnal salivary cortisol levels, among a sample of 51 children and adolescents with ASD and a control group of typically developing participants matched on age, gender, and pubertal status. The results revealed that typical diurnal patterns were

evident in both groups. There was a small effect of autism diagnosis on cortisol levels, with individuals diagnosed with autism presenting with higher cortisol levels than their typically developing peers, although this finding was not statistically significant. Among participants with autism, cortisol levels were not differentiated by age (i.e., child or adolescent), gender (i.e., male or female), verbal ability (i.e., vocal verbal or non-vocal verbal), presence or absence of co-morbid intellectual disability, or psychotropic medication usage (i.e., use or non-use).

This study also examined the relationship between physiological stress (average daily cortisol levels), parent-reported stress, and teacher- or parent-reported engagement in repetitive, stereotyped behaviour, self-injury, and aggressive behaviour among 61 children and adolescents with ASD. Parent-reported stress was not found to be associated with physiological stress. Further, there was no discernible correlation between measures of parent reported- or physiological stress and engagement in challenging behaviour for the sample as a whole. However, participants engaging in low levels of stereotypy presented with lower cortisol levels (large effect) than participants engaging in high levels of stereotypy, although this difference was statistically non-significant which may be related to the small sample size for these comparisons. These results suggest that stereotypy may be an indicator of stress among persons with ASD, but that it does not appear to function as a coping mechanism or stress-reducing mechanism.

Study 3

Study 3 (Lydon et al., 2015c/Chapter 4) examined HR patterns surrounding the various topographies of motor stereotypy exhibited by five children and adolescents diagnosed with autism in the context of a functional assessment. This study identified little correspondence between HR and stereotypy for four of the five participants. For the additional participant, findings suggested that stereotypy may have been negatively reinforced by a subsequent decrease in HR that was consistently associated with

occurrences of motor stereotypy. These results contrast with previous research findings in this area. Further, findings of a lower level of HR during distress, as compared to elation, among three of the five participants were also potentially suggestive of physiological blunting in response to stress among some individuals diagnosed with autism.

Study 4

Finally, Study 4 (Chapter 5) employed measures of HR and EDA in the context of a brief functional analysis of the repetitive, stereotyped behaviours of six individuals with autism. Similar to Study 3 (Lydon et al., 2015c/Chapter 4), the results revealed little association between physiological activity and engagement in repetitive, stereotyped behaviours among participants. However, as observed in Study 3 (Lydon et al., 2015c/Chapter 4), an atypical physiological response to stress that was possibly indicative of physiological blunting was noted for two of the children who took part in this study.

Implications of the Research Findings

Clinical Utility of Psychophysiological Assessments of Challenging Behaviour

Romanczyk and Matthews (1998) have previously noted the conceptual validity of psychophysiological assessments of challenging behaviour, such as those described in Chapters 3-5 of the current research program, but noted that the clinical utility of such assessments remains to be demonstrated. Similarly, other scholars (e.g., Cohen et al., 2011; Langthorne et al., 2014; Langthorne et al., 2007; Langthorne et al., 2008; Symons & Roberts, 2013; Youngstrom & Reyes, 2015) have also called for an examination of the most useful means of incorporating technological advances and physiological measurement into behavioural frameworks or paradigms, and the determination of the incremental validity offered by psychophysiological assessment in the context of standard clinical assessments. Given the mixed outcomes of Studies 2-4, the true clinical utility of such assessments for persons with autism remains unclear. A relationship between physiological arousal and challenging behaviour was identified in Studies 2 and 3 for

some, but not all, participants. This outcome suggests that there is merit in such analyses but that additional research is required to identify the characteristics of participants or challenging behaviour for which such assessments will be most useful and effective. For example, it is possible that such assessments may be more useful among lower functioning individuals or with challenging behaviours that are solely maintained by automatic sources of reinforcement. It is imperative that future research establishes the individuals for whom these assessments may be recommended; such assessments have the potential to identify an array of effective treatment options such as arousal-inducing or arousal-decreasing strategies that would otherwise not be considered. However, these assessments, and the analysis of the resulting physiological data, are a time-consuming affair that, given the conflicting findings observed among participants in this research, may not be recommended for universal use. Youngstrom and Reyes have previously noted that in order for psychophysiological assessment to "earn a place in the clinical toolkit" (2015; p.357) that it must be demonstrated to offer significant incremental validity over standard assessment protocols, especially given the greater intrusiveness of such assessment techniques. Thus, in addition to determining the parameters in which psychophysiological assessments of challenging behaviour may be recommended, future research that examines methods in which such assessments can be refined and improved for optimal efficaciousness and efficiency would be of use. The current research program has presented protocols for incorporating physiological measures alongside functional assessments and during functional analyses, and for analysing the resulting data, but it is possible that researchers may identify more efficient and effective means of doing this in the future.

Future Directions in the Extension of an Operant Account of Challenging Behaviour

As described in Chapter 1, an abundance of theoretical work, emerging as early as the 1960s, has posited a link between physiological activity or reactivity and engagement

in challenging behaviour by persons with ASD. However, there are a number of limitations to the various theories that may impede research and knowledge development in this field. These theories vary in their propositions but the majority center on the idea of some underlying dysregulation in physiological functioning amongst persons with ASD. These theories have suggested, for example, impairments in autonomic regulation that result in physiological hyper- or hypo-arousal that impacts upon overt behaviour (Hutt et al., 1964; Hutt & Hutt, 1968; Sugarman et al., 2014) or in circadian rhythms (Tordjman et al., 2015). Such hypotheses may be considered problematic as research concerning physiological functioning in ASD is highly inconsistent (Lydon et al., in press/Study 1: Chapter 2; Nuske et al., 2014; Taylor & Corbett, 2014; Tordjman et al., 2014) and has failed to yield any consistent evidence regarding specific physiological abnormalities or deficits to support these suggestions. In this way, data heretofore has failed to support the fundamental suggestions of these theories (i.e., the physiological deficit suggested to contribute to engagement in the behaviour has not been demonstrated to exist), calling into question the validity and utility of these theories. Further, there is also an unfortunate disconnect between the theoretical work and the empirical research in this area (see Chapter 1); few of the individuals who have proposed theories linking physiological activity and challenging behaviour have also conducted empirical investigations in this area. As a result, the studies that have been conducted have typically not been designed to test any specific theory and there has been no progress towards refuting, validating, or improving upon the existing theoretical underpinnings. In addition to these failings, there is also a demonstrable lack of consideration in the existing theoretical work of what is known about challenging behaviour and why it may occur. For example, the decades of theoretical work and empirical research on operant theory that have contributed to applied behaviour analysis' success in identifying the maintaining variables for challenging behaviour and effective strategies for reducing and eliminating such behaviours, have not considerably impacted

theoretical directions. While some of the theoreticians (e.g., Guess & Carr, 1991; Groden et al., 1994; Langthorne et al., 2007; Langthorne et al., 2008; Langthorne et al., 2014; Lydon et al., 2013a; Romanczyk et al., 1986) have couched their writings in behaviour analytic terms, and within a behaviour analytic account, this is relatively rare. Given the dominance of operant theory in explaining behaviour, failure to consider this account may be considered detrimental to both the theory on the potential association between physiological activity and challenging behaviour, and to the empirical studies conducted to assess for a possible association between these variables.

Such discussion prompts reflection on the relative lack of involvement of behaviour analysts and behavioural researchers in this area. Anecdotally, it may be noted that the dissemination of this research program at behaviour analytic conferences was sometimes met with hostility. Prolific authors in the field of ABA expressed the opinion that it was not the domain of behaviour analysts to be concerned with what was happening internally during instances of challenging behaviour, that the concept of automatic reinforcement did not warrant further exploration or clarification, and that in cases where the identification of a behaviour as being automatically reinforced did not contribute to the development of an effective antecedent- or reinforcement-based intervention, the utility of punishment-based procedures could be employed. Other behaviour analysts (e.g., Baum, 2011) have also strongly repudiated the importance or appropriateness of the study of internal processes or events, such as physiological activity, by behaviour analysts in the research literature. This resistance appears to stem from the conceptualisation of physiological activity as an internal event, private event, or covert behaviour, and the consideration of these as unnecessary or inappropriate sources of data for behaviour analysts, or at odds with behavioural theory. It may be suggested that this stance could be considered improvident. The lack of acknowledgement, study, and consideration of covert behaviour in behavioural theory and research has been criticised in a number of texts (e.g., Donohue & Palmer,

1994; Stemmer, 2003; Tourinho, 2006). Skinner (1965) referred to private events as those "distinguished by [their] limited accessibility but not, so far as we know, by any special structure or nature" (p. 257). This quotation could be interpreted as suggesting that, if accessible, these events may warrant consideration by behaviour analysts. While private events have received relatively limited attention in the behavioural literature, Skinner's definition, characterised by the inaccessibility of these events, has persisted and has been used as the basis for the discrimination between public and private events, and also to define those variables which are considered to be relevant to behaviour analysis. However, Donohue and Palmer (1994) have emphasised that "the observability of a response is not determined by its intensity or magnitude, but by the characteristics or tools of the observer. … We must avoid the temptation to think of covert behavior as a kind of behavior, with properties essentially different from overt behavior. Rather, all behavior lies on a continuum of observability" (p. 275). Similarly, Skinner seemed to emphasise the important role of these events in behaviour, referring repeatedly in his writings to the "world within the skin" (1973, p.24), nothing that "an adequate science of behavior must consider events taking place within the skin of the organism... It can deal with these events without assuming that they have any special nature or must be known in any special way" (1969, p. 228), and emphasisising that the ability to measure physiological activity could only contribute to making "the picture of human action more nearly complete" (1974, p.125). Skinner also described specific situations in which physiological events or activity could alternatively elicit overt behaviour, reinforce overt behaviour, or act as a discriminative stimulus for overt behaviour. Therefore, it may be suggested that the increasing observability or accessibility of physiological activity, stemming from recent advances in technology, now offers behaviour analysts and behavioural researchers the opportunity to investigate the relationship between physiological activity and behaviour, a potential for the advancement of the science of behaviour that was recognised decades ago

by Skinner. Indeed this may promulgate a paradigm shift whereby observation and analysis of physiological activity as “private events” becomes part of the assessment and treatment of challenging behaviour in this population. Research programs such as that reported here may thus be considered appropriate means of inquiry for behaviour analysts and may contribute to an improved understanding of behaviour for typically developing persons and persons with psychiatric or developmental disorders.

The potential for psychophysiological measurement and assessment to contribute to behavioural theory and understanding is perhaps greatest for the concept of automatic reinforcement. The current research program offered the potential to elucidate the internal stimulation responsible for the maintenance of challenging behaviour identified as automatically reinforced. The utility and veracity of the concept of automatic reinforcement has frequently been called into question by researchers; Barrera et al. (2007, p. 30) argue that the concept of automatic reinforcement is flawed because of “how unknown, invisible, fiction, and perhaps unfalsifiable” the consequences it refers to are. Similarly, Vaughan and Michael (1982) contend that without evidence or proof of automatic reinforcement, operant theory is incomplete and argue against the acceptance of “hypothetical constructs as explanations” (p. 218) by behaviour analysts. Recently, Hagopian and colleagues (2015) have described three subtypes of automatically reinforced SIB and identified differences in functional analysis outcomes, treatment outcomes, presence of other challenging behaviour, and the behavioural function of other challenging behaviours. One of the subtypes described includes SIB that is insensitive to the environment, resistant to reinforcement-based interventions, and suggested by the authors to have a biological basis. Hagopian et al. emphasise the need for biobehavioural research in order to further our understanding of such automatically reinforced behaviour. Past research on automatic reinforcement processes in the developmental disability population has been constrained due to the difficulty, or impossibility, of assessing and measuring the

internal stimulation hypothesised to play a role in these behaviours. Recent advancements in technology mean that, now, it may be possible to begin to empirically investigate at least some of the cases in which behaviours are suggested to be automatically reinforced by interoceptive stimuli. Previous theoretical and empirical work has suggested that levels of, or changes in, physiological arousal may provide the internal stimulation responsible for the perpetuation of automatically reinforced challenging behaviour (Barrera et al., 2007; Freeman et al., 1999; Gabriels et al., 2013; Groden et al., 1994; Guess & Carr, 1991; Hirstein et al., 2001; Hoch et al., 2010; Hoch et al., 2013; Hutt et al., 1964; Hutt et al., 1968; Hutt et al., 1975; Jennett et al., 2011; Lewis et al., 1975; Lydon et al., 2013; Moskowitz et al., 2013; Romanczyk, 1986; Romanczyk & Matthews, 1998; Sroufe et al., 1973; Sugarman et al., 2014; Symons et al., 2003; Symons et al., 2011; Verhoeven et al., 1999; Willemsen et al., 1998; Young & Clements, 1975). The current research program adds some support to the suggestion that this may be true for some, but not all, individuals with ASD who engage in automatically reinforced challenging behaviour. Research focused on subtyping challenging behaviours, such as that conducted by Hagopian et al. (2015) with stereotyped SIB, may help with the identification of the individuals for whom such psychophysiological assessments may be useful. In the current study, it is perhaps unsurprising that specific sources of internal stimulation may play a role in the maintenance of challenging behaviour in some individuals with ASD; for example, previous research has also shown that other forms of sensation such as visual or auditory stimulation are responsible for the maintenance of topographies of challenging behaviour among individuals with autism (e.g., Rincover et al., 1979; Tang et al., 2003). Future research may continue to capitalise upon advances in technology for measuring both brain and physiological activity in order to further explicate and validate automatic reinforcement processes in autism and move towards a more nuanced understanding of automatically reinforced challenging behaviour.

Stress in Autism

Study 2 (Lydon et al., 2015b/Chapter 3) investigated stress among children and adolescents with autism and matched typically developing peers. There were no significant differences in cortisol levels between the groups, although a small effect of diagnosis was observed. This outcome is interesting given that individuals with autism are considered to have a unique vulnerability to stress (Groden et al., 1994), and that high rates of anxiety disorders exist in this population (Simonoff et al., 2008). It may be considered concerning that parent-reported stress was not associated with children's actual physiological stress in Study 2 (Lydon et al., 2015b/Chapter 3), suggesting that individuals with autism may experience high stress without their primary carers or other individuals in their environment being aware of this. Such findings suggest that more frequent use of physiological monitoring of individuals with ASD may be considered in order to better understand their experiences and levels of stress, and to identify those in need of specifically tailored treatment protocols. A number of interventional strategies exist that may help with the reduction of stress in this population including social skills training interventions (Cotugno, 2009; Solomon, Goodlin-Jones, & Anders, 2004), directly trained self-management techniques, cognitive behaviour therapy, psychoeducation, and relaxation training (Groden et al., 1994; Ooi et al., 2008; Tsai, 2006). Future research may aim to evaluate effective and feasible means of screening or assessing for stress in this population and subsequently providing effective treatment to those individuals with autism who present with elevated levels of stress.

Stress and Stereotypy

The relationship between stress and stereotypy observed in Study 2 (Lydon et al., 2015b/Chapter 3) is an important finding of the current research program, although it may be considered preliminary given the statistical non-significance of this finding in spite of the large effect size observed. There is a prevailing belief that stereotyped behaviour is

adaptive for persons with autism and may function as a coping mechanism for stress (Groden et al., 1994). The only previous study (Gabriels et al., 2013) that has assessed the relationship between cortisol and repetitive behaviours in autism was in concordance with this suggestion. The perception of stereotypy as adaptive may lead to an acceptance of such behaviours and non-intervention by practitioners, in spite of the reported negative impact on academic and social engagement (Koegel & Covert, 1972; Koegel et al., 1974; Lovaas et al., 1971). Such a perception may be considered problematic given that the findings of the current study appear to suggest the opposite; that stereotypy is an indicator of stress but does not function as a coping mechanism. The conflicting findings in this area highlight the need for future investigation of the relationship between stereotypy and stress, and also highlight the importance of conducting behavioural assessments of stereotyped behaviour, in an attempt to understand the function or meaning of such behaviours for individual children and adolescents with autism. Future research may also aim to examine the impact of stress-management interventions on repetitive behaviours among individuals with autism for whom behavioural assessments have failed to identify the maintaining source of reinforcement for these behaviours.

Desynchrony between Physiological Activity and Observed Behaviour

A number of findings of the current research program were suggestive of desynchrony between physiological activity and overt behaviour among the participants with autism. These included the lack of correlation between PR and behaviour topographies in a number of studies reviewed in Study 1 (Lydon et al., in press/Chapter 2), the lack of correspondence between parent-reported stress and physiological stress in Study 2 (Lydon et al., 2015b/Chapter 3), a finding also observed in previous research studies (Bitsika et al., 2014; Bitsika, Sharpley, Andronicos, & Agnew, 2015; Corbett et al., 2006), and the low levels of physiological activation during overt distress observed for some participants in Studies 3 (Chapter 4) and 4 (Chapter 5). These findings are

disquieting as, given the communication deficits associated with ASD, there is often a high degree of reliance upon overt behaviour and affect, or parental report, to infer wellbeing and mood, reactions to environmental events, and emotional state among children and adolescents with autism. The results of the current research program suggest that the assumption that overt behaviour and affect reflects internal activation and emotion is false and may lead to incorrect perceptions of internal state. This is problematic as it may lead to the non-recognition of what could be harmful physiological or emotional states, such as high levels of stress. Other researchers (e.g., Lyons, Walla, & Arthur-Kelly, 2012) have also described the difficulties in accurately understanding the internal states and experiences of children with disabilities and the importance of this knowledge for assessing subjective wellbeing and quality of life. Physiological assessment methods have the potential to contribute to an improved understanding of the experiences of persons with disabilities (Boltë, Golan, Goodwin, & Zwaigenbaum, 2010; Lyons et al., 2012). Technological advances have made this a feasible suggestion and research such as that reported here has demonstrated that monitoring of physiological activity can contribute to a better understanding of the experiences of children with autism (Hedman et al., 2012). Thus, future research should take caution when using such indirect measures and perhaps consider optimal means of including physiological monitoring within the lives of children and adolescents with autism in order to facilitate better understanding of internal states.

Interventions based on the Assumption of a Relationship between Physiological Arousal and Challenging Behaviour

Our findings also highlight that caution is necessary in the implementation of interventions predicated on an assumed relationship between physiological arousal and challenging behaviour. Interventional strategies for challenging behaviour such as the widely adopted Low Arousal Approach (for a review, see: McDonnell et al., 2015) propose that physiological arousal is an important determinant of challenging behaviour and

suggest interventional techniques based on this assumed relationship. For example, McDonnell et al. (2015) call for the development of low arousal environments, and use of low arousal responses as a standard reaction to challenging behaviour (i.e., the reduction of demand, avoidance of eye contact, whispered communication etc.). If such interventions are not based on a prior functional anlaysis of the problem behaviour(s), they may have the potential to negatively reinforce escape-maintained problem behaviour and lead to an increased frequency or intensity of such behaviour. They may also impede the utilisation of appropriate function-based behavioural interventions, despite research highlighting the greater efficacy of interventions based upon identified behavioural function (Campbell, 2003; Heyvaert et al., 2014; Kahng et al., 2002). The current program of research has demonstrated both the feasibility of assessing the association between these variables, and the falsity of the assumption that challenging behaviour is universally associated with physiological arousal among individuals diagnosed with autism. While arousal-inducing strategies such as physical exercise (see: Lang et al., 2010; Kasner et al., 2012) and arousal-reducing strategies such as the low arousal approach (McDonnell et al., 2015) and relaxation techniques (Mullins & Christian, 2001; To & Chan, 2000) have previously been found to be effective in the treatment of challenging behaviour, we suggest that the demonstration of a relationship between physiological arousal and the targeted challenging behaviour is necessary prior to their implementation. This will indicate the likely effectiveness of such interventions with individuals to whom they are suited as well as guiding the most appropriate manner of implementation.

Recommendations for Future Research

In addition to the suggestions for future research that have been made throughout this chapter, it is also possible to recommend a number of additional avenues for future research exploration that would greatly contribute to our understanding of the experiences

of persons with ASD, physiological functioning in this population, and the relationship between physiological activity and challenging behaviour.

Physiological profiles of Autism

The heterogeneity of persons diagnosed with autism is well recognised (e.g., Allen et al., 2013; Stagg et al., 2013) and is considered to contribute to the highly inconsistent findings of many research studies examining physiological activity among persons diagnosed with autism (e.g., Benevides & Lane, 2013; Klusek et al., 2014; Lydon et al., in press/Study 1: Chapter 2; Taylor & Corbett, 2014). London (2014) has described the many deficits of DSM diagnoses including the lack of information pertaining to the aetiology of disorders, the lack of biomarkers for disorders, the lack of distinction between many DSM disorders, and the simplistic descriptions of symptoms. London emphasises that the categorical diagnosis of autism is misguided and that researchers should seek to devise other means through which persons with autism can be sub-grouped in order to better conduct research within this population. Studies reviewed in Study 1 (Chapter 2) that were suggestive of the existence of sub-groups of individuals with ASD with differing physiological profiles (e.g., Hirstein et al., 2001; Schoen et al., 2008), the inconsistency in outcomes among studies reviewed in Study 1 (Chapter 2), and the lack of correspondence between the findings of Chapters 3, 4, and 5 and previous research studies in this area, highlight the need for more useful methods of categorisation alongside the diagnosis of ASD. For example, if individuals with autism can be grouped into hypo-aroused, normally aroused, and hyper-aroused groups based upon their baseline physiological activity and patterns of PR, then it could be recommended that these groups be studied independently as increased and decreased PR have been linked to differential health outcomes (e.g., Lovallo, 2011), and baseline physiological activity has been implicated as a determinant of treatment outcome (Stadler et al., 2008). It is also reasonable to suggest that the relationship between physiological arousal and challenging behaviour may differ according

to the individuals' physiological profile and future research that subdivides individuals with ASD according to physiological profiles may further contribute to our understanding of this relationship between these variables. Thus, it is recommended that future research seek to investigate and define differential physiological profiles within the ASD population and to examine the implications of these profiles for important variables relating to ASD including challenging behaviour, health outcomes, treatment outcomes, and functional outcomes. In concordance with this suggestion, Klusek and colleagues (2014) have recently emphasised that "deconstructing clinical groups at the biological level" (p. 141) may be a key means of improving the accuracy and reliability of the diagnosis of ASD.

Physiological Blunting

The results of Studies 3 (Chapter 4) and 4 (Chapter 5) were tentatively suggestive of physiological blunting among a number of the participating children and adolescents with autism. Physiological blunting was suggested by the observation of lower levels of HR during overt distress as typically distress is associated with elevated HR, similar to the HR elevations during elation that were observed. As the current research study was not designed to assess physiological stress responses, these suggestions must be considered tentative and require further research and empirical validation. Physiological blunting has been identified in other disorders including depression, ADHD, and addictive disorders (Paris et al., 2010; Pesonen et al., 2011; Schwerdtfeger & Rosenkaimer, 2011) and has been linked to a myriad of negative outcomes among typically developing persons including engagement in challenging behaviour (Bauer et al., 2002). The research of physiological blunting is in its infancy among typically developing persons (Lovallo, 2011) and, to my knowledge, the phenomenon has not yet been investigated among persons with autism. However, the study of physiological blunting in this population may be recommended given the negative health outcomes associated with this style of stress responsivity (Lovallo, 2011). Further research is therefore required to establish whether

physiological blunting is prevalent among individuals with autism, to examine ways in which this may be adaptive for persons with autism, and to assess the short- and long-term implications of this for these individuals.

Investigation of Physiological Functioning among Lower-functioning Individuals with Autism

Study 1 (Lydon et al., in press/Chapter 2) revealed that a preponderance of research has examined PR among individuals with high-functioning ASD, but that individuals with low-functioning ASD have received relatively little attention in these research studies. This finding is likely attributable to the greater ease of including individuals without a co-occurring intellectual disability in such experiments. However, researchers and reviewers have suggested previously that cognitive ability impacts on physiological activity and responses (e.g., Kootz et al., 1982; Putnam et al., 2015; Taylor & Corbett, 2014; Tordjman et al., 2014), although this is not always evident among psychophysiological studies of individuals with ASD (Nuske et al., 2014). Technological advances have resulted in lightweight and unobtrusive physiological monitoring devices and made the inclusion of lower functioning individuals with ASD in psychophysiological studies increasingly possible (Porges, 2013; Symons & Roberts, 2013). The inclusion of participants with autism and co-occurring intellectual disabilities in the three experimental studies detailed in this dissertation highlights the feasibility of their participation in studies using a variety of physiological measures and data collection protocols. Future research must thus endeavour to include lower-functioning individuals with ASD in psychophysiological research studies in order to determine the impact of level of functioning on physiological activity, and to better explore physiological activity and reactivity among a greater representation of persons with autism.

Limitations

This program of research had a number of strengths including the diversity of the individuals with autism who participated, the adherence to best practice for the inclusion of participants with ASD in the three psychophysiological experiments conducted (Kylliainen et al., 2014), the range of topographies of challenging behaviours with which participants presented, the collection of data in participants' natural settings, and the development of a novel measure of methodological quality for use with quasi-experimental, non-interventional, psychophysiological research studies that may aid future researchers to plan and conduct high-quality research studies in this area. However, in spite of these considerable strengths, the current research also had a number of important limitations that must be considered when interpreting its findings.

Sampling

The reliance upon convenience sampling of individuals with autism in the current research program may be considered a key limitation. In all three experimental studies, the criteria for participant inclusion were minimally restrictive. The difficulties of participant recruitment precluded the utilisation of more stringent inclusion criteria as was initially intended. Our participant pool was also restricted by the necessity that participants could tolerate the saliva collection procedure (Lydon et al., 2015b/Study 2: Chapter 3) or wearing the physiological recording devices (Studies 3 and 4: Chapters 4 and 5 respectively). This requirement led to the exclusion of a number of children and adolescents who presented with high levels of sensory sensitivity, showed distress during the habituation or data collection phases, or who engaged in aggressive behaviours that would have endangered those responsible for the collection of saliva samples or for the application and management of the physiological recording devices. However, it is important to note that the reliance upon convenience sampling may have impacted upon the outcomes of these studies and may limit the external validity of the results. For example, different results may

have been obtained had participants in Study 2 (Lydon et al., 2015b/Chapter 3) been recruited based upon their engagement in either “low” or “high levels” of stereotypy, SIB, and aggressive/destructive behaviour. Similarly, for Study 3 (Lydon et al., 2015c/Chapter 4), the recruitment of participants whose stereotypy was insensitive to sources of social reinforcement, as per the QABF, or for whom engagement in motor stereotypy was associated with overt signs of reduced or elevated physiological arousal may have produced different experimental outcomes. The more stringent inclusion criteria employed in Study 4 (Chapter 5) were an attempt to remediate the deficits of Study 3 (Lydon et al., 2015c/Chapter 4) by requiring participants’ challenging behaviours to be either maintained primarily by automatic reinforcement or resistant to previous behavioural intervention, increasing the likelihood that there was a physiological component to the behaviour. Future research may wish to utilise more stringent inclusion criteria when researching the relationship between challenging behaviour and physiological arousal in autism.

Sample Size

The sample sizes of Studies 2-4 may also be considered a limitation of the current research program. In Study 2 (Lydon et al., 2015b/Chapter 3), a small effect of diagnosis on stress, and a large effect of engagement in stereotypy and its relation to stress, were identified but these differences were not statistically significant which may indicate an underpowered analysis. However, cortisol collection and analysis is widely recognised to be an expensive endeavour (Adam & Kumari, 2009) and, in the current research program, insufficient financial resources were available to test a larger sample. However, the sample of individuals with autism who did participate in this research study nonetheless compares favourably to previous similar research studies (e.g., Gabriels et al., 2013; Symons et al., 2003; Symons et al., 2011; Verhoeven et al., 1999). The sample size of Studies 3 and 4 was determined based on previous research studies in this area that have successfully identified relationships between physiological arousal and challenging behaviours with

smaller sample sizes (e.g., Barrera et al., 2007; Freeman et al., 1999; Lydon et al., 2013). However, it is possible that the inclusion of greater numbers of participants in these studies may have impacted upon experimental outcomes or aided with the determination of characteristics of the individual or their challenging behaviour that are indicative of a relationship between challenging behaviour and physiological activity. Future research incorporating a greater number of participants may thus yield a considerable understanding of the link between challenging behaviour and physiological arousal in autism.

Participant Characterisation

This research may also be criticised for its reliance upon participants' extant psychological records, completed by independent psychologists, for the determination of participants' primary and co-morbid diagnoses and any existing medical conditions. As a result of this, participants were diagnosed with autism using a variety of diagnostic instruments and it was not possible to determine the reliability of diagnosis. All participants had psychological reports that were deemed of sufficient rigour and quality to justify receipt of ASD-specific services by the Irish Department of Education and Skills responsible for special education in Ireland. However, given the varying comprehensiveness of participants' psychological reports, and the duration since the most recent psychological assessment, it is possible that participant characterisation may have been deficient in some cases. However, it was necessary to rely upon participants' psychological records given the large number of participants in Study 2 (Lydon et al., 2015b/Chapter 3), and the limited time for which participants in all studies were available to the researchers. Future research may aim to assess participant characteristcs more thoroughly, to verify the accuracy of diagnoses, and to characterise level of intellectual functioning more comprehensively. Such participant information may aid with the identification of mediators of atypical physiological functioning, or any relationship between challenging behaviour and physiological activity.

Missing Physiological Data

An additional limitation of the current dissertation was the proportion of missing physiological data observed. In Study 2 (Lydon et al., 2015b/Chapter 3), missing cortisol data arose from a failure to collect a sufficient volume of saliva for analysis but this was an infrequent issue (<5% of missing samples). However, there was a notably higher proportion of missing HR data in Studies 3 and 4, and EDA data in Study 4. Missing HR data was typically due to malfunctioning equipment that may have resulted from a number of challenges in working with this population. Notably, the primary cause of malfunction of the HR and EDA monitors was the movement of the recording device by participants. This occurred at a high frequency for participants who engaged in challenging behaviour that involved the movement of the torso, or hand to chest movements. The proportion of missing physiological data observed is not unique to the current study; many of the studies reviewed in Study 2 (Lydon et al., 2015b/Chapter 3) reported missing data and this is also frequent among studies assessing the relationship between physiological activity and challenging behaviour (e.g., Hoch et al., 2013; Moskowitz et al., 2013; Romanczyk & Matthews, 1998). Further, Kylliainen and colleagues (2014) have highlighted the near-impossibility of completely avoiding technical problems when employing measures of physiological activity with children diagnosed with autism, and emphasised that the exclusion of participants due to missing data is inadvisable. For this reason, all participants were retained, but occurrences of challenging behaviour for which the corresponding physiological data was missing were excluded from all analyses.

Data Analysis Methodologies

The methods of data analysis employed in Studies 3 and 4 (Chapters 4 and 5 respectively) may also be critiqued. The analysis of high volumes of data originating from a single individual is highly complex and was given much thought and consideration in the current research program. Throughout this research program, we have collected the data

best suited to addressing our research questions and applied best judgement to analysing these data. This approach was considered preferable to collecting simple data analysable via orthodox statistical techniques but data that did not adequately address our research questions. It has previously been suggested that research studies examining the relationship between physiological arousal and challenging behaviour should employ statistical techniques such as time-series analysis and multi-level modelling (Cohen et al., 2011). However, the current program of research sought to develop practical protocols for the collection of physiological data in the context of functional assessment and analysis and methods of data analysis that were applicable to, and useful for, practicing behaviour analysts and clinicians. Previous research has shown that the use of complex statistical techniques in research can isolate clinicians and practitioners (e.g., Cohen et al., 1986; Herbert, 2003; Stewart & Chambless, 2010; Stewart et al. 2012) and hinder their understanding of research findings. Thus, this research program aimed to employ methods of data analysis appropriate for the examination of our research question that were also straightforward and could be employed by practitioners without a high degree of statistical knowledge.

Non-provision of Intervention

Finally, the current study could be faulted for the non-provision of intervention to participants for whom a relationship between physiological arousal and challenging behaviour was identified. The implementation of an appropriate intervention in such cases, and assessment of its efficacy, would have allowed for the confirmation of a functional relation between the two variables. For example, in Study 2 (Lydon et al., 2015b/Chapter 3), participants who engaged in high levels of stereotypy and who were identified as having significantly higher cortisol levels than their peers who did not engage in high levels of stereotypy, could have been provided with an intervention intended to reduce stress; reduction of environmental demand, identification and alleviation of stressors,

functional communication skills to express stress or feelings of discomfort, and the teaching of relaxation skills may all have been effective strategies. The implementation of such an intervention and the assessment of its effects on engagement in stereotypy would have allowed for a stronger demonstration of a relationship between the two variables. Similarly, in Studies 3 and 4, the implementation of appropriate arousal-reducing or arousal-increasing interventions and the evaluation of their impact upon the frequency of engagement in challenging behaviour would have yielded stronger support for the association between physiological activity and behaviour. However, there were a number of reasons such interventions were not evaluated in the current research program. Key among these was the passage of time between data collection and the completion of physiological analysis. In many cases, participants had graduated from, or transferred services, by the completion of data collection for the study. Further, most participants were attending schools or services with limited personnel resources which hampered the implementation of behavioural interventions.

Conclusion

The current research program used both group and single-subject research designs to examine the contribution of psychophysiological measurement to our understanding of physiological activity in autism, and its association with challenging behaviour. The findings suggest that psychophysiological assessments among persons with autism have some value and can contribute to behaviour analysts' understanding, assessment, and treatment of challenging behaviour among persons with autism. Such assessments may also yield an improved understanding of the experiences and wellbeing of individuals with autism. However, there are caveats to these assertions. The research reported in this dissertation indicates that such psychophysiological assessments may not be useful for all persons with autism, and that future research is required to develop guidelines or parameters for the optimal utilisation of these assessments. Further research is also

warranted to determine the most effective manner of utilising advances in technology to improve the understanding of autism and the complex co-occurring behaviours that are associated with the condition. Investigation of potential physiological sub-groups among persons with autism may yield a better understanding of physiological activity and reactivity in this population, and may elucidate the relationship between physiological activity and challenging behaviour, a question that is increasingly important in the field of ABA.

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Appendix A

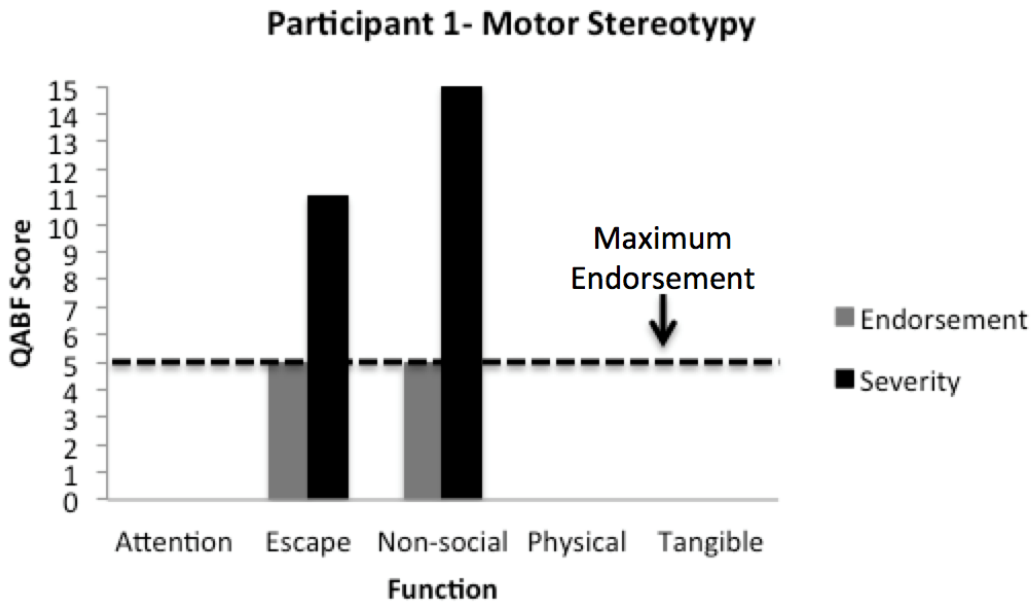

Figure A.1 QABF scores for Participant 1's motor stereotypy.

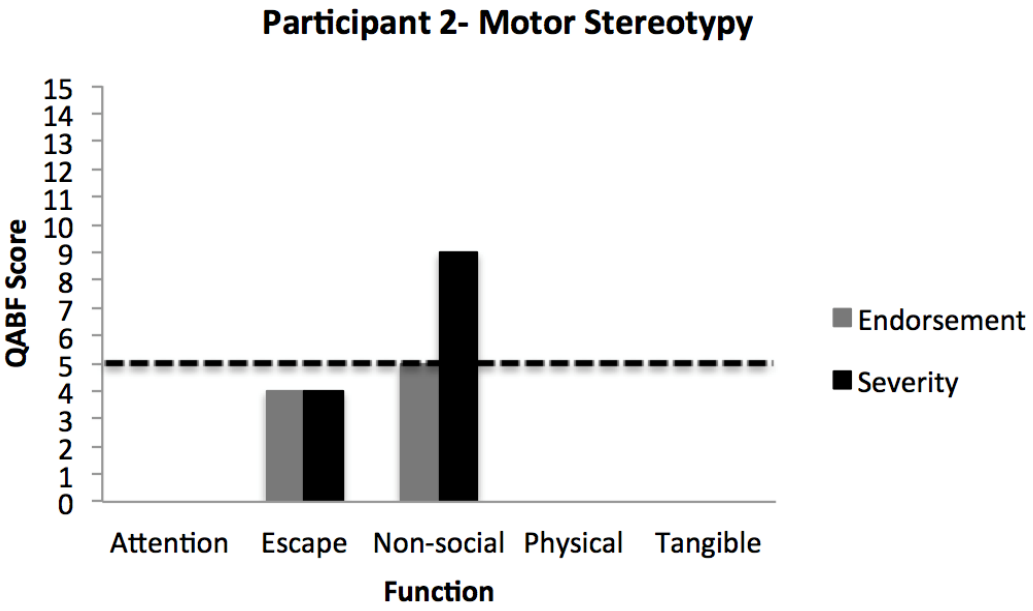

Figure A.2 QABF scores for Participant 2's motor stereotypy.

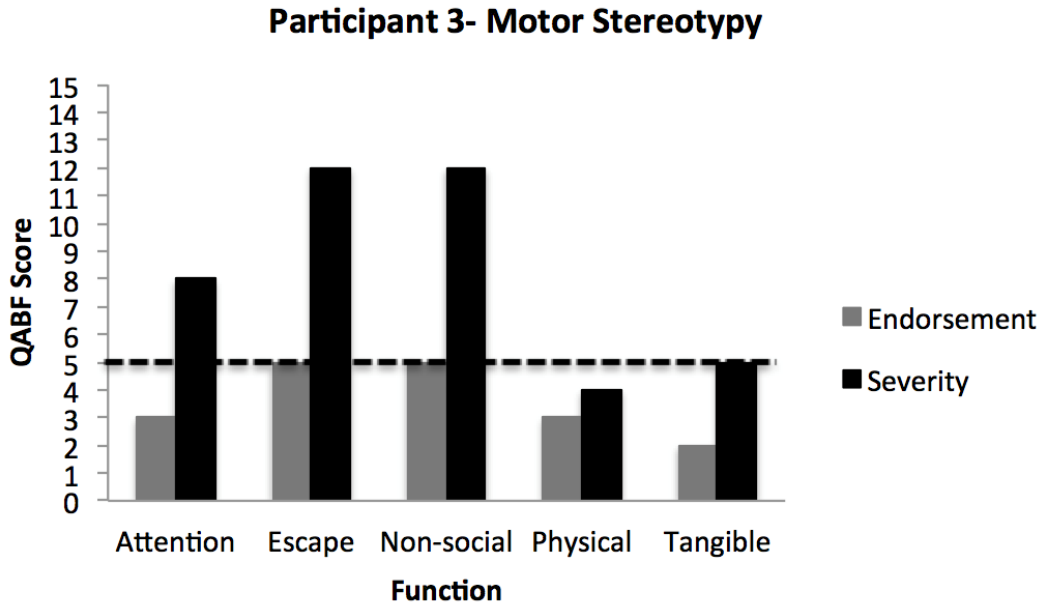

Figure A.3 QABF scores for Participant 3's motor stereotypy.

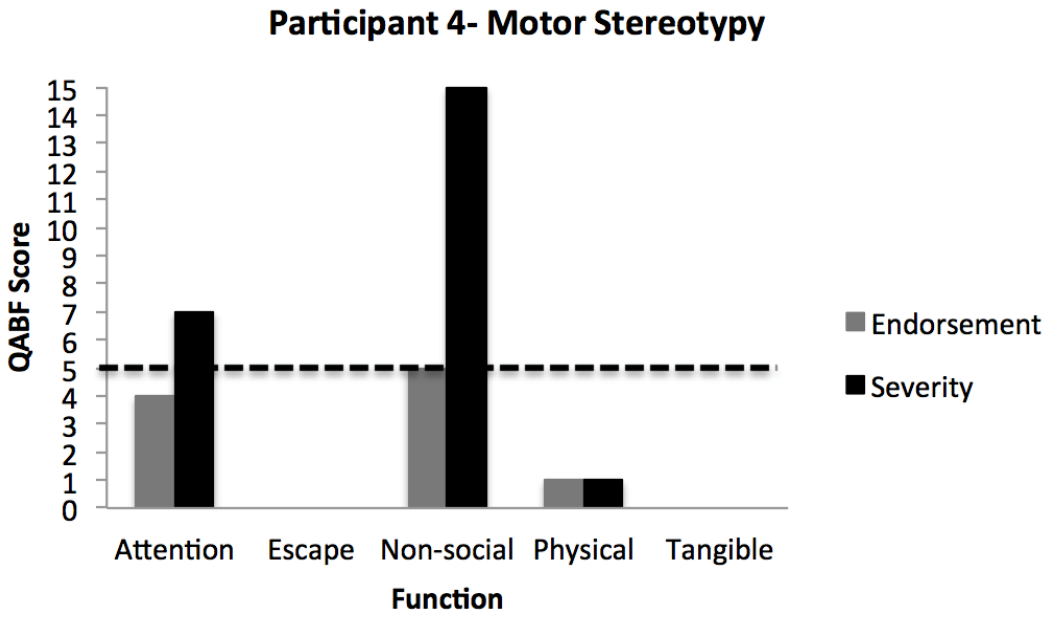

Figure A.4 QABF scores for Participant 4's motor stereotypy.

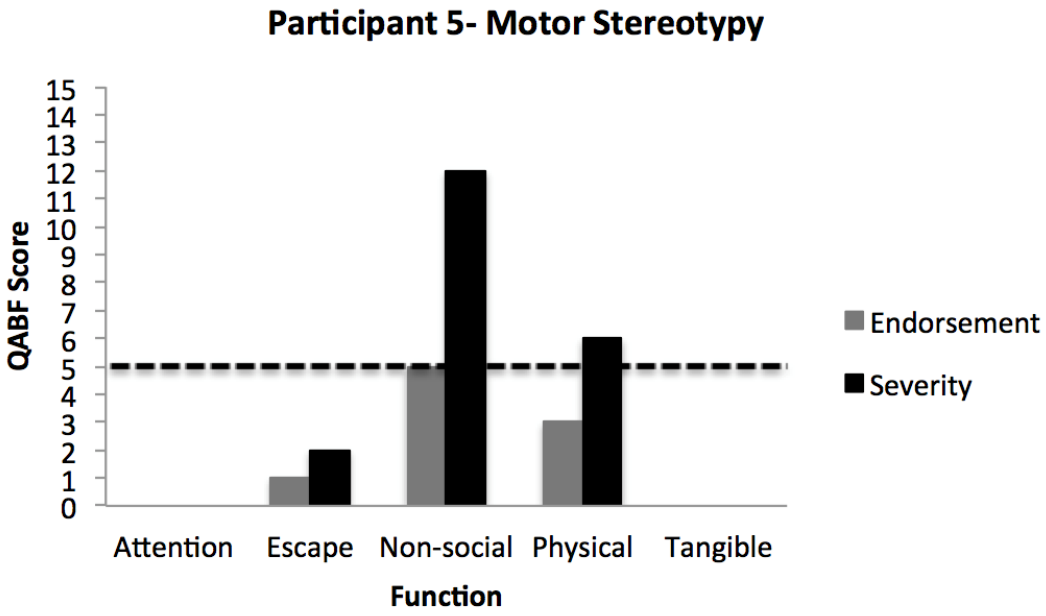

Figure A.5 QABF scores for Participant 5's motor stereotypy.

Appendix B

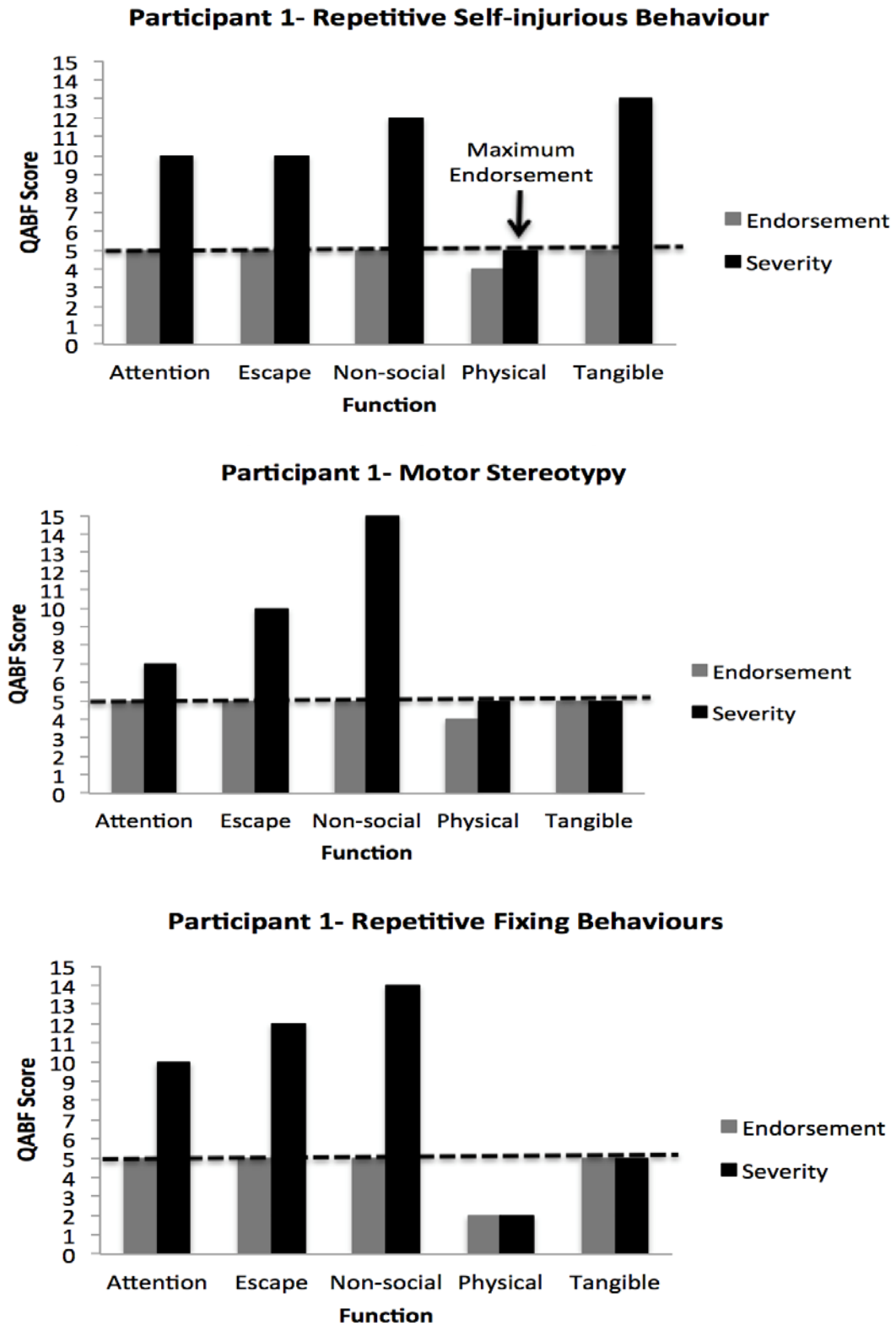

Figure B.1 QABF scores for Participant 1's repetitive self-injurious behaviours (upper panel), motor stereotypy (middle panel), and repetitive fixing behaviours (lower panel).

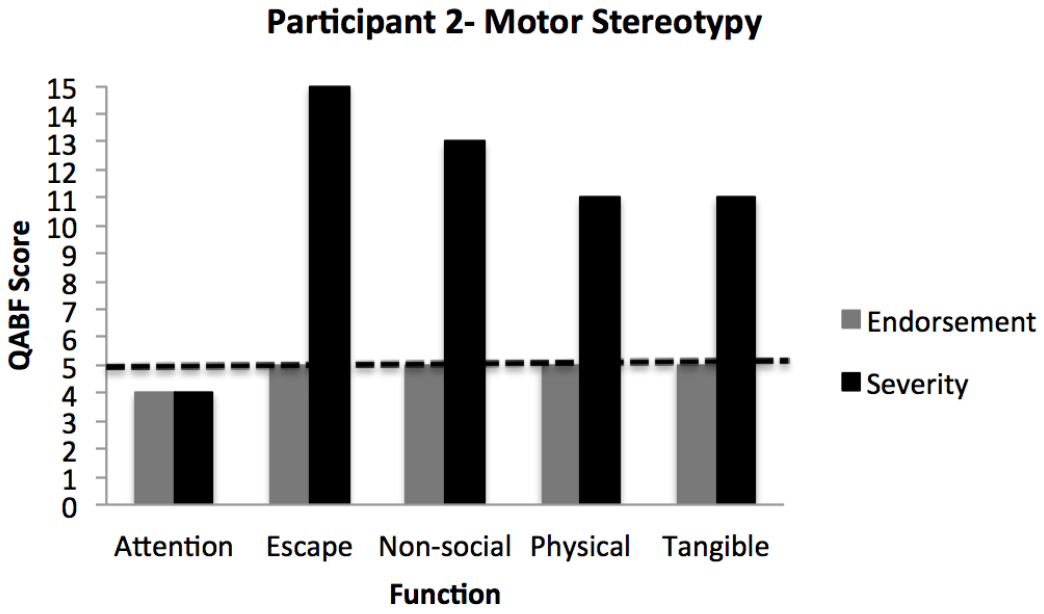

Figure B.2 QABF scores for Participant 2's motor stereotypy.

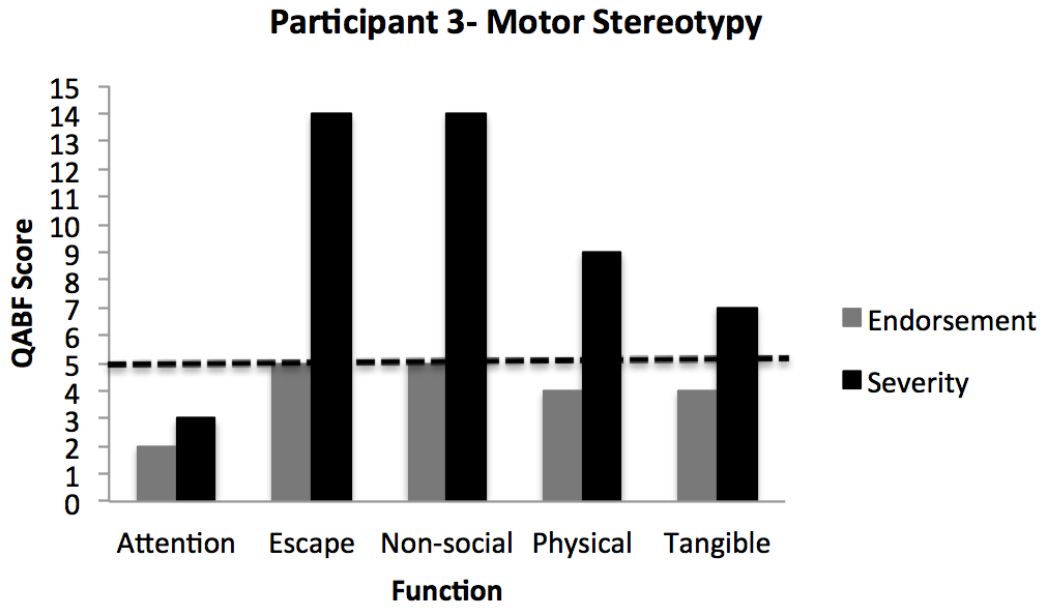

Figure B.3 QABF scores for Participant 3's motor stereotypy.

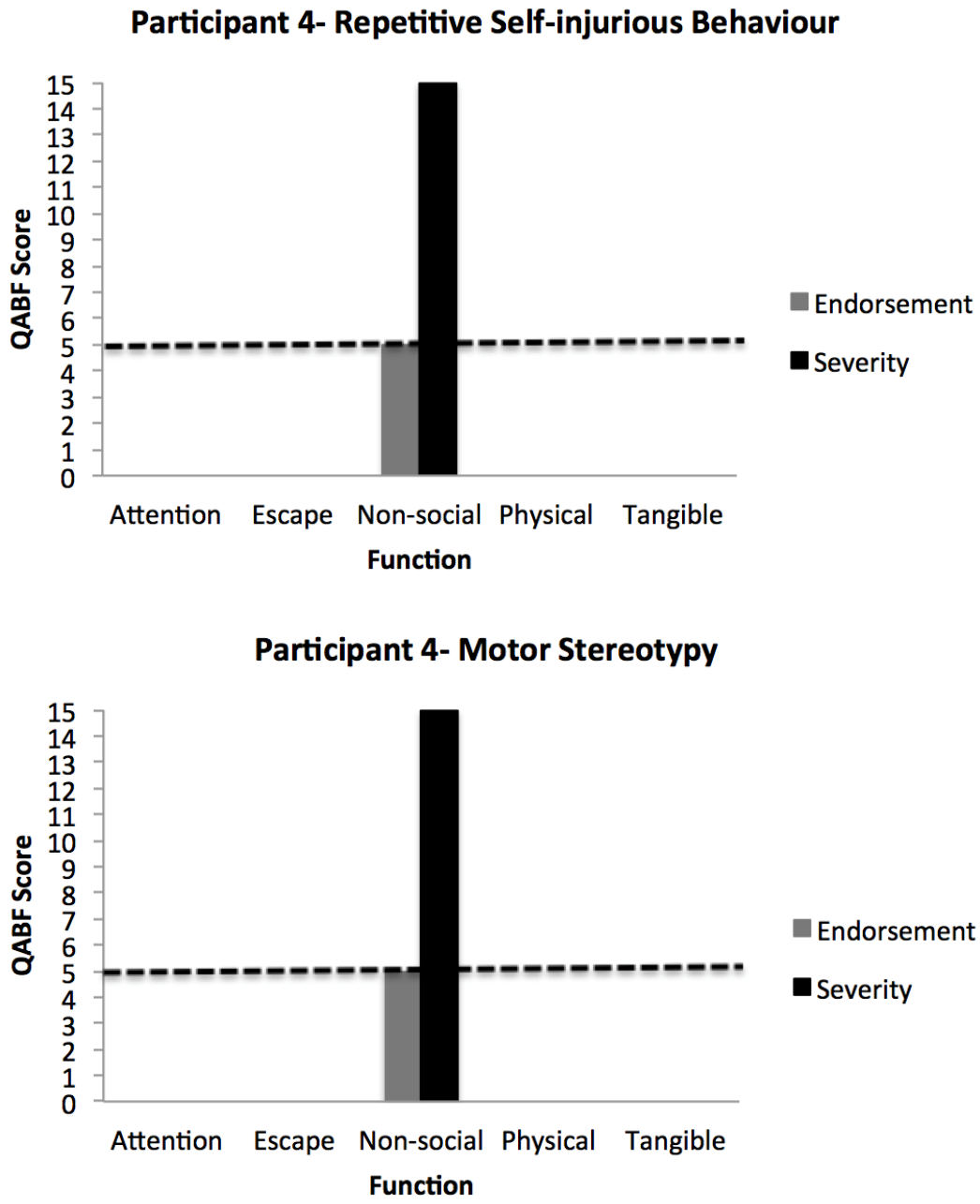

Figure B.4 QABF scores for Participant 4's repetitive self-injurious behaviours (upper panel), and motor stereotypy (lower panel).

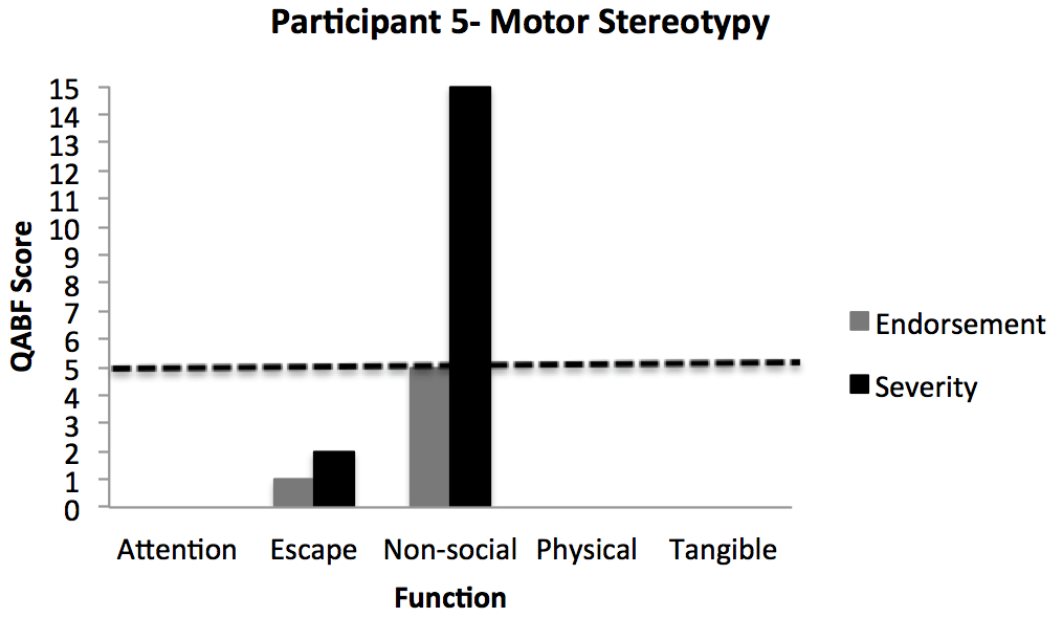

Figure B.5 QABF scores for Participant 5's motor stereotypy.

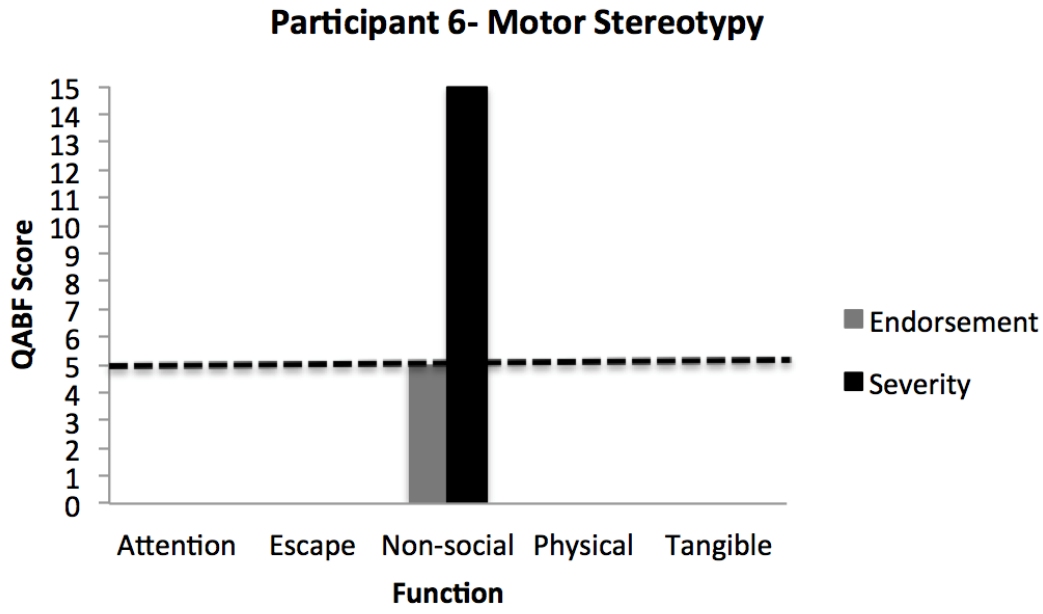

Figure B.6 QABF scores for Participant 6's motor stereotypy.