School of Psychology and Speech Pathology

Music, Arousal and Self-Injurious Behaviour: A Three-Stage Mediating Model for Children with Low Functioning Autism

Jeremy Kent Marriott

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DECLARATION

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. The proposed research study received human research ethics approval from the Curtin University Human Research Ethics Committee (EC00262), Approval Number HR138/2011.

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ABSTRACT

Reports suggest that listening to calming music can reduce arousal. Reports also suggest that reducing arousal can reduce self-injurious behaviour (SIB). Prior to the present research, these reports had not been investigated amongst school-aged boys with autism spectrum disorder (ASD) and an intellectual disability (ID) known as low functioning autism (LFA). The following research investigated the potential for biological arousal to mediate a relationship between music listening and SIB amongst school-aged boys with LFA.

Study 1 incorporated musical factors, form, segment and expert musical consultations into the selection of a calming music playlist. Thirty-two primary carers of males with LFA rated the calming properties of 6 two-minute musical segments based on how calming they would be for their son with LFA. A generalised linear mixed model (GLMM) identified the second movement of Ludwig van Beethoven's sonata No. 8 in C minor, Op. 13 adagio cantabile known as his *Sonata Pathetique* and the first movement of Beethoven's sonata no. 14 en do dièse mineur, Op.27, No. 2 adagio sostenuto known as his *Moonlight Sonata* as significantly more calming than the other segments in the playlist but not more calming than each other. Beethoven's *Sonata Pathetique* was then selected via expert musical consultation for application in Study 2 and Study 3.

Study 2 assessed whether school-aged boys with LFA who listened to Beethoven's *Sonata Pathetique* would record lower biological arousal after being exposed to an adapted trier social stress test for children (TSST-C) in the form of a morning school bus ride simulation. Fifteen boys with LFA were randomly allocated to a music listening group and 15 boys, matched by school-age and LFA diagnosis, were randomly allocated to a no music group. Saliva samples were collected from the boys before, and at 4 time-points after the simulation. The samples were analysed for biomarkers of arousal: salivary cortisol (sCort) concentration as a hypothalamic-pituitary-adrenocortical (HPA) axis biomarker and salivary alpha-amylase (sAA) activity as an autonomic nervous system (ANS) biomarker. The music group recorded significantly lower sCort across the collection time-points compared with the no music group. This result suggested that music could reduce arousal for boys with LFA in a controlled and demanding environment.

A single-case design comprising two conditions with repeated trials, Study 3 assessed the sCort, sAA and SIB frequencies of three school-aged boys with LFA ranging in age from 14 to 16 years (M = 15.3 years, SD = 1.1 years), on the same day each week over four consecutive weeks. Two of the boys (named Bill and Harry) exhibited ear-blocking SIB and one (named George) with pica SIB. Treatment Day 1 and Day 3, administered during weeks one and three, required the boys to take their usual morning school bus ride, provide saliva samples across five time-points and have video footage of their behaviour recorded by the researcher. Treatment Day 2 and Day 4, administered during weeks two and four, were identical to Day 1 and Day 3 with the addition of the *Sonata Pathetique* for the final 14 minutes and 35 seconds of the morning bus ride. The saliva samples were analysed for sCort and sAA as per Study 2. In addition, video footage collected on the school bus and at school was analysed for SIB frequencies using a partial interval recording (PIR) analysis. Motivation Assessment Scale (MAS) ratings provided by primary carers were recorded then analysed to identify possible SIB motivating factors.

Study 3 assessed the validity of seven hypotheses using two distinct statistical stages. Stage 1 tested Hypotheses 1 to 3 and stage 2 assessed Hypotheses 4 to 7. Hypothesis 1 aimed to determine if listening to calming music could reduce the frequency of SIB (ear-blocking or pica) compared to non-exposure. Hypotheses 2 and 3 assessed whether listening to calming music was associated with reductions in sCort concentrations and sAA activity respectively when compared non-exposure. Hypotheses 4 and 5 aimed to determine if reductions in sCort and sAA would be associated with reductions in SIB frequencies respectively. Hypothesis 6 assessed if changes in sCort were associated with changes in sAA. Lastly, Hypothesis 7 assessed if increases in SIB frequencies were associated with either a symmetrical association or an asymmetrical association between sCort and sAA (Bauer et al., 2002).

Hypotheses 1 through 3 were tested using a conventional single-case design visual analysis. Hypotheses 4 through 7, used an unconventional single-case analytic technique by conducting series of Fisher's Exact tests to determine potential associations between sCort concentration, sAA activity and SIB frequencies. sCort concentration and sAA activity means and standard deviations (*SDs*) for each participant were converted to z-scores and analysed across the four treatment days of the study. SIB frequency data were gathered via 10-second PIRs as rated by the

researcher and two independent inter-raters based on video footage recorded on the bus and at school.

In relation to Bill and Harry, stage 1 of the analysis revealed no support for Hypotheses 1, 2, or 3. Further, stage 2 of the analysis revealed no support for Hypotheses 4, 5, 6, or 7. In relation to George, listening to calming music reduced the frequency of his pica SIB on Day 2 (Music); supporting Hypothesis 1. However, this was not replicated at Day 4 (Music). The remainder of George's stage 1 analysis revealed no support for Hypotheses 2, or 3. In addition, his stage 2 analysis revealed no support for Hypotheses 4, 5, 6, or 7.

Of interest, the significant sCort reduction detected in the controlled school bus simulation environment of Study 2 did not generalise to the naturalistic morning school bus ride setting of Study 3. Key findings, theoretical implications, clinical implications, strengths, limitations and recommendations for future research are discussed.

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LIST OF ABBREVIATIONS

ASD	autism spectrum disorder
ID	intellectual disability
LFA	low functioning autism
SIB	self-injurious behaviour
FA	functional analysis
MAS	motivation assessment scale
TSST	trier social stress test
TSST-C	trier social stress test for children
sCort	salivary cortisol
HPA	hypothalamic-pituitary-adrenocortical axis
sAA	salivary alpha-amylase
ANS	autonomic nervous system
PIR	partial interval recording

CHAPTER 1: INTRODUCTION

Children with autism spectrum disorder (ASD) and an intellectual disability (ID) have a diagnosis known as low functioning autism (LFA; Cheung, Chan, Sze, Leung, & To, 2010). Unfortunately, up to 40 percent people with LFA are vulnerable to exhibiting self-injurious behaviour (SIB; Bodfish et al., 1995; Horner, Carr, Strain, Todd, & Reed, 2002; Kahng, Iwata, & Levin, 2002).

SIB is complex, physically damaging, life-threatening and occurs in various forms (Campbell, 2003; Fee & Matson, 1992; Minshawi, 2008; Novak, 2003; Tate & Baroff, 1966). For example, ear-gouging/ear-blocking SIB, occurs when fingers are used to damage the skin and structure of the ear (Iwata, Dorsey, Slifer, Bauman, & Richman, 1994). Often exhibited by people with LFA, pica SIB occurs when inedible objects are placed onto the lips or inside of the mouth repetitively (Lai, Lombardo, & Baron-Cohen, 2014; Singh & Winton, 1984). Pica can result in lead poisoning and require surgery to remove objects that have been inadvertently ingested (Matson, Belva, Hattier, & Matson, 2011). The occurrence of SIB can have negative impacts on school, social and developmental progress for children with disabilities such as LFA (Glasson et al., 2008; Machalicek, O'Reilly, Beretvas, Sigafoos, & Lancioni, 2007; Matson, Sipes, Fodstad, & Fitzgerald, 2011; McClintock, Hall, & Oliver, 2003; Richman, 2008). Without sufficient quantities of empirical research and evidence-based interventions for SIB, the prognosis for children can be dire (Campbell, 1995).

Treating SIB can be challenging (Barrera, Violo, & Graver, 2007; Horner et al., 2002; Iwata, Dorsey, et al., 1994). As a result, the peer-reviewed behavioural and pharmacological treatments designed to treat repetitive behaviour such as SIB exhibited by children with ASD are lacking in both number and efficacy (Boyd, Woodard, & Bodfish, 2013). However, it has been hypothesised that SIB may have an underlying biological component (Canitano & Scandurra, 2011). More specifically, there may be a link between SIB and increasing levels of arousal (Jennett, Hagopian, & Beaulieu, 2011). Interestingly, people with ASD can respond well to music and this may reduce arousal and externalising behaviours such as SIB for those with communication difficulties such as those with LFA (Ford, 1999; Papagiannopoulou, 2015).

Therefore, the purpose of the following research was to assess three separate but interconnected components of a theoretical three-stage mediating model to determine if biological arousal could mediate a relationship between music listening and SIB among school-aged boys with LFA. Chapter 2 defines, identifies and highlights relevant literature in the areas of ASD, LFA, SIB, music therapy, biological arousal, research methods and models of child biology/behaviour. Chapter 3 provides an evidence-based rationale for the administration of the three separate but interconnected studies associated with the model. Chapter 4 explains the design and implementation of Study 1, which identified a calming musical segment for use in Study 2 and Study 3. Chapter 5 describes how Study 2 used the music selected in Study 1 to assess a potential link between music listening and a reduction in biological arousal among 30 boys with LFA before and after being exposed to a demand condition in the controlled environment of a morning school bus ride simulator. Chapter 6 explains how Study 3 assessed the potential for a reduction in biological arousal to mediate a relationship between music listening and a reduction in SIB frequencies among boys with LFA in the naturalistic setting of an actual morning school bus ride; ultimately testing the validity of the theoretical three-stage mediating model. Lastly, Chapter 7 discusses the results of all three studies and offers suggestions for future research.

CHAPTER 2: LITERATURE REVIEW

2.1 Autism Spectrum Disorder from Past to Present

The first known attempts at defining what is now known as autism spectrum disorder (ASD) were published in the 1940's by Leo Kanner and Hans Asperger. In *Autistic disturbances of affective contact*, Kanner described the social, communicative, sensory and behavioural "peculiarities" of eight boys and three girls under 12 years of age (Henninger & Taylor, 2013; Kanner, 1943, p. 217; Lai et al., 2014). Overall, these features were defined as constant, persistent, pervasive and "extremely autistic" (Kanner, 1943, p. 222). Of the 11 cases presented in this seminal ASD publication, the case of five year-old Donald T. was the most extensively described.

Donald T. was reported by Kanner to exhibit social skill, gesture and reciprocity deficits with specific and restricted areas of interest, behavioural repetitiveness, and sensory likes and dislikes (Lai et al., 2014). His speech was described as echolalic and repetitive and he insisted on categorising objects into shapes and colours. His bodily movements were described as repetitive and stereotyped and he relied heavily on rules, schedules and literal verbal comprehension. He had difficulties maintaining attention and transitioning between tasks. Socially, Donald T. was described as an egocentric boy who had little interest in others and displayed infrequent periods of direct eye-to-eye gaze. In addition to these difficulties, Kanner reported that Donald T. possessed above-average memory and learning abilities (Kanner, 1943). Importantly, Kanner's detailed case presentation of Donald T. was one of the first published attempts to define the features of childhood ASD.

A year after Kanner's paper was published, Hans Asperger published *Autistic psychopathy in childhood*. Originally published in German, this paper was translated into English by Uta Frith in 1991. In this translated paper, Asperger described the communicative, social, behavioural and physical features of four children as "particularly interesting" (Asperger, 1944 as cited in Frith, 1991, p. 37). Further, and with similar detail to the case presentation of Donald T., Asperger published the case of a "highly unusual boy" he named as Fritz V. (Asperger, 1944 as cited in Frith, 1991, p. 39). Six-year-old Fritz V. was reported to be devoid of love, empathy and affection whilst being hyperactive and impulsive with social difficulties, echolalic speech and a lack of interest in others (Lai et al., 2014). His behaviour was described as combative, avoidant, non-compliant and socially inappropriate. He was reported to have deficits

in fine and gross motor skills. His speech rate, volume and rhythm, social play, direct eye-to-eye gaze, body posture and physical features were described as "odd" (Asperger, 1944 as cited in Frith, 1991, p. 42). He was reported to have stomach issues as a result of licking and eating pencils, paper and other objects with high frequency and in large quantities. Fritz V. had learning difficulties, restrictive and specific areas of interest with unusual and stereotyped body movements. In an attempt to define the origin of his features, Asperger suggested that his presentation may be the result of encephalitis or a juvenile form of schizophrenia. In addition to the aforementioned difficulties and deficits, Asperger reported that Fritz V.'s speech, memory, and mathematical abilities were "unusual" in that they were significantly better than what would be expected of his same-aged peers (Asperger, 1944 as cited in Frith, 1991, p. 44). The case presentation of Fritz V. served a similar purpose to the case of Donald T in that it was also one of the initial attempts to describe the features of childhood ASD.

A combination of the cases reported by Kanner and Asperger, especially those of Donald T. and Fritz V., were the catalyst for the subsequent 30-years of discussions, investigations and intrigue regarding this undefined condition (Baker, 2013). For example, in the 1950's and 1960's, children presenting with features similar to those of Donald T. and Fritz V. were suggested to have childhood schizophrenia. Without the interest and intrigue created by the seminal works of Kanner and Asperger, more formal attempts to diagnostically conceptualise the condition may have never prevailed (Lai et al., 2014).

The third edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-III) included the first official diagnostic criteria to define the features of what was known as *infantile autism* (American Psychiatric Association, 1980; Reschke-Hernández, 2011). Comprised of six criteria, infantile autism was defined by atypical responses to social situations and environmental stimuli with severe communication deficits; all of which needed to emerge before 30 months of age (American Psychiatric Association, 1980; Baker, 2013; see Appendix A). The criteria were located within *disorders usually first evident in infancy, childhood, or adolescence* and categorised as a *pervasive developmental disorder* (American Psychiatric Association, 1980).

The inclusion of infantile autism in the DSM-III further heightened clinical interest, intrigue and academic inquiry into this highly heterogeneous condition (Baker, 2013). As a result, when the DSM-III was revised in 1987, the DSM-III-R included an expanded version of the diagnostic criteria and renamed it *autistic disorder*

(Baker, 2013) within the Pervasive Developmental Delay Not Otherwise Specified (PPD-NOS) classification. This expansion saw the diagnostic criteria increase in number from six to 16 and categorised these into three as opposed to a single domain: social deficits, communication difficulties and restricted activities and interests (American Psychiatric Association, 1987; see Appendix B).

The DSM-IV consolidated the diagnostic criteria for autistic disorder by reducing the criterion in number from 16 to 12 and distributing them equally across three domains with more specific descriptions: social deficits; communicative difficulties; and restricted, repetitive and stereotyped patterns of behaviour (American Psychiatric Association, 1994; see Appendix C). Further, autistic disorder became classified as one of the five *pervasive developmental disorders* in addition to Rett's disorder, Childhood Disintegrative Disorder, Asperger's Disorder and PDD-NOS. The diagnostic title, criteria, and classification for autistic disorder remained mostly unchanged when the DSM-IV-TR was published (American Psychiatric Association, 2000; see Appendix D). Further, pervasive developmental disorders could also be defined as autism spectrum disorders and PDD-NOS included a specifier known as atypical autism.

The DSM-IV-TR enabled diagnosticians to specify the level of functional deficit associated with autistic disorder features. For example, people meeting the diagnostic criteria with a low level of daily, behavioural and cognitive deficits were deemed to have high functioning autism (HFA). The converse was specified for those with Low Functioning Autism (LFA). By the time the DSM-IV-TR was published, autistic disorder had emerged as the most prevalent of the *neurodevelopmental disorders* (American Psychiatric Association, 2000; Cheung et al., 2010; Matson & Nebel-Schwalm, 2007).

The most recent edition of the DSM, the DSM-5 published several changes to the diagnostic criteria (see Appendix E). Firstly, *autism spectrum disorder* (ASD) became the diagnostic term used to represent previous DSM-IV TR diagnoses of autistic disorder, childhood disintegrative disorder, Asperger's Disorder and PDD-NOS. As such, the diagnostic criteria for ASD represented a spectrum of presentations. Secondly, the DSM-5 enabled diagnosticians to report the severity of ASD symptoms to indicate the level of support required. Thirdly, the number of diagnostic criteria and domains were reduced from 12 criteria across three domains in the DSM-IV-TR to seven criteria across two domains in the DSM-5. As part of this revision, the domain

which had previously defined specific communication deficits in the DSM-IV-TR was removed. ASD was classified as a *neurodevelopmental disorder* with deficits of social interaction, communication and restricted and repetitive behaviours (American Psychiatric Association, 2013).

Parallel to, and as a result of the seminal works by Kanner and Asperger combined with the diagnostic criteria revisions, the body of knowledge associated with ASD has grown significantly over the last 70 years (Lai et al., 2014). Now, more than ever, ASD is known as a disorder of vast heterogeneity, complexity, functional differences and severities with an ever increasing prevalence (Hall & Kelley, 2014).

2.2 ASD Prevalence: Global, National and Local

Estimating the global prevalence of ASD, as is for any disorder, a difficult task (Mandell & Lecavalier, 2014). However, reports suggest that the prevalence of ASD in the United States of America (USA) has increased consistently over the last three decades; with a noticeable acceleration in younger children (Camarata, 2014; Samadi, Mahmoodizadeh, & McConkey, 2012). For example, ASD prevalence estimates amongst eight year-old children in the USA has risen from one in every 110 births in 2006, to one in every 88 births in 2008 and to one in every 68 births in 2010 (Centers for Disease Control and Prevention, 2009, 2012, 2014). Globally, ASD prevalence rates are reported to range from 20 (Fombonne, 2009) to 62 in every 10,000 births (Elsabbagh et al., 2012).

The *Autism in Australia* report published by the Australian Bureau of Statistics (ABS) estimated that 0.5 percent of the total Australian population has a diagnosis of ASD (Australian Bureau of Statistics, 2012). A recent study published by Randall et al. (2016) reported the prevalence of ASD amongst 4 to 5-year-old Australian children to be 1.5%. Specific to the location of the present research, the number of people diagnosed with ASD in Western Australia (WA) has risen consistently between the years 1999 to 2005 (Glasson et al., 2008). Specifically, the state-wide prevalence rate of ASD in WA is estimated to be 0.4 percent (Australian Bureau of Statistics, 2015). The increasing prevalence rates of ASD globally, nationally and locally have resulted in a greater need to identify its causal factors.

2.3 ASD Causal Theories and Factors

Historically, causal theories associated with ASD have been numerous and contentious. In the 1950's and 1960's, ASD was thought to have been the result of emotionally unresponsive "refrigerator" mothers (Baker, 2013, p. 1090; Yudell,

Tabor, Dawson, Rossi, & Newschaffer, 2013). In the late 1990's, Wakefield et al. (1998) suggested that ASD may be caused by the measles mumps rubella (MMR) vaccine. This theory was systematically investigated, comprehensively refuted and eventually retracted (Baird et al., 2008; Doja & Roberts, 2006; Madsen et al., 2002; Mrozek-Budzyn, Kieltyka, & Majewska, 2010; Richler et al., 2006; Smeeth et al., 2004; Tannous et al., 2014).

Currently, the DSM-5 identifies ASD as having genetic and environmental causal risk factors (American Psychiatric Association, 2013). Contemporary ASD casual risk factor research has begun to focus on identifying non-specific and specific risk factors. Non-specific risk factors include an older parental age at birth and low birth weight (American Psychiatric Association, 2013).

Specific risk factors include a family history of ASD, genetic disorders and mutations and environmental toxins such as pesticides and pollutants (Baker, 2013; Dawson, 2008; Posey, Stigler, Erickson, & McDougle, 2008). Specific risk factor research estimates the incidence of ASD in twin's ranges from 37 to more than 90 percent (Geschwind, 2011). Further, microarray research reports that some copy-number variations may be linked to the incidence of ASD (Hall & Kelley, 2014). In addition, biomarker research has reported that approximately 80% of mothers with the thyroid peroxidase antibody (TPO-Ab+), a marker of autoimmune thyroiditis, give birth to children who are subsequently diagnosed with ASD (Brown et al., 2015).

So informative is this specific risk factor research approach that millions of dollars in the USA have been allocated to the associated field of *epigenetics* (Camarata, 2014; Lai et al., 2014). Epigenetic specific risk factor research investigates the potential for environmental change to modify gene expression without altering DNA sequences and has resulted in the identification of genes associated with Rett's Syndrome, Fragile X Syndrome and Tuberous Sclerosis (Hall & Kelley, 2014). Despite these findings in other fields, specific risk factors associated with ASD are not yet conclusive (Kidd et al., 2012; Lai et al., 2014; Posey et al., 2008; Yudell et al., 2013). What is known, however, is that people with ASD can present with a spectrum of abilities. As such, people with ASD who also have an intellectual disability (ID), known as Low Functioning Autism (LFA), are most severely affected (Cheung et al., 2010). LFA occurs in approximately 45 percent of those diagnosed with ASD (Lai et al., 2014).

2.4 Defining Low Functioning Autism

Just as the diagnostic criteria for ASD have evolved, so have the diagnostic criteria for ID. The first edition of the DSM, the DSM-I published by the APA in 1952 used the term *mental deficiency* to described what is known today as ID (American Psychiatric Association, 1952; see Appendix F). In 1952, this condition was described as an "impairment of brain tissue" caused by familial and hereditary factors (American Psychiatric Association, 1952, p. 86). By the time the DSM-II was published, the diagnostic term changed to *mental retardation* and was defined as "subnormal general intellectual functioning", emergent during the early developmental period and was thought to be linked to social, learning and/or maturity maladjustments (American Psychiatric Association, 1968, p. 14; see Appendix G). The DSM-III retained this diagnostic term, however required those diagnosed to record both significantly below average intelligence quotient (IQ) and adaptive functioning abilities which had emerged prior to 18 years of age (American Psychiatric Association, 1980; see Appendix H). Few changes in the diagnostic criteria for mental retardation occurred when the DSM-III-R, DSM-IV and DSM-IV-TR were published (American Psychiatric Association, 1987, 1994; 2000; see Appendices I, J & K). However, the DSM-5 revised the diagnostic term to that of *intellectual disability* (ID) and defined it as a condition which emerged in the early developmental period and featured deficits in both intellectual and adaptive functioning (American Psychiatric Association, 2013; see Appendix L). Further, a diagnosis of ID required the examinee to record a full scale intelligence quotient (FSIQ) score of 70 plus or minus 5 points or below on a standardised cognitive assessment (American Psychiatric Association, 2013). In addition to the DSM-5, the American Association on Intellectual and Developmental Disabilities (AAIDD) adds that people with an ID are significantly limited in both intelligence and the ability to adapt to daily, social and practical demands (American Association on Intellectual and Developmental Disabilities, 2013).

In combination, the features of ASD and ID, as is the case for those with LFA, can result in the presentation of extremely challenging behaviours. In the case of children with LFA, the most serious and frequently occurring of these challenging behaviours is known as self-injurious behaviour (SIB; American Psychiatric Association, 2013; Horner et al., 2002; Matson & Nebel-Schwalm, 2007; McClintock et al., 2003; Minshawi, 2008).

2.5 Self-Injurious Behaviour: A Definition

Self-injurious behaviour is a complex, auto-aggressive, self-destructive and severe behaviour with numerous types (see Table 2.1; Campbell, 2003; Minshawi, 2008; Novak, 2003; Tate & Baroff, 1966). It can be broadly defined as any forceful contact to the face, chest, or abdomen with a body part of varied frequency and intensity, resulting in "physical injury to the individual's own body" (Healey, Ahern, Graff, & Libby, 2001; Matson & LoVullo, 2008; Tate & Baroff, 1966, p. 281; Wachtel et al., 2009).

SIB can range in severity from mild/occasional to severe/frequent and be exhibited by people with ID and a range of other syndromes such as Lesch-Nyhan syndrome, Prader-Willi syndrome, borderline personality disorder and Cornelia de Lange syndrome (Fee & Matson, 1992; Hersen & Sturmey, 2012; Winchel & Stanley, 1991). In addition, SIB is commonly exhibited by people with ASD and is estimated to occur in up to 40% of those with LFA (Bodfish et al., 1995; Canitano & Scandurra, 2011; Kahng, Iwata, & Lewin, 2002; Lai et al., 2014; Novak, 2003). Indicatively, Smith, Press, Koenig, and Kinnealey (2005) reported that up to 160,000 people in the USA with developmental disabilities such as LFA exhibit destructive behaviour such as SIB at an annual treatment cost of more than $3 billion. The term SIB will refer to children with LFA in the present research as opposed to terms such as non-suicidal self-injury or deliberate self-harm that refers to people with a range of psychological and/or psychiatric conditions (Plener et al., 2015).

SIB has particular clinical relevance to therapists striving to help children with living with LFA (American Psychiatric Association, 2013). For example, Bishop, Richler, and Lord (2006) found that between 21.2 and 57.0 percent of 830 children living with ASD and pervasive developmental disorders in the USA engaged in SIB. In addition, the prevalence of SIB amongst children living in France with ASD was reported to be up to 50 percent, with 14.6 percent of these exhibiting severe SIB (Baghdadli, Pascal, Grisi, & Aussilloux, 2003).

School-aged children with ASD who exhibit SIB can experience frustration, attention difficulties, low academic achievement with limited adaptive, self-help and social skill development (Ford, 1999; Gorman-Smith & Matson, 1985; Matson, Mahan, Hess, Fodstad, & Neal, 2010). If SIB persists through childhood into adulthood, it can result in significant tissue damage including deep lacerations, muscular injuries, bone fractures, hearing and vision losses as well as physical

disfigurements, infections, brain injury and even death (Hersen & Sturmey, 2012; Lai et al., 2014; Matson & LoVullo, 2008; Minshawi, 2008; Novak, 2003; Wachtel, Jaffe, & Kellner, 2011). Further, it is not uncommon for primary carers to be tasked with physically restraining those with LFA to prevent tissue damage when serious injury appears imminent (Lai et al., 2014). Because of the potential physical and psychological harm associated with SIB, effective treatment is essential.

Table 2.1

Types of Self-Injurious Behaviour

SIB	Description
Ear-pulling and gouging	Using fingers or fingernails to pull or dig at ears (Iwata, Dorsey, et al., 1994).
PICA	Placing inedible and non-nutritional objects onto lips or inside mouth (Singh & Winton, 1984).
Head-banging	Forceful contact made to the head (Davenport, Lutz, Tiefenbacher, Novak, & Meyer, 2008; Iwata, Dorsey, et al., 1994; Kurtz et al., 2003; Novak, 2003; Vollmer, Iwata, Zarcone, Smith, & Mazaleski, 1993).
Head-hitting	The forceful contact made by body parts with any portion of the head (American Psychiatric Association, 2013; Ford, 1999; Iwata, Dorsey, et al., 1994; Kurtz et al., 2003; Vollmer et al., 1993).
Self-biting	Biting any part of the body inclusive of wrists (Davenport et al., 2008; Iwata, Dorsey, et al., 1994; Kurtz et al., 2003; Novak, 2003).
Hair-pulling	Pulling hair away from the head with force (Davenport et al., 2008; Iwata, Dorsey, et al., 1994; Kurtz et al., 2003).
Teeth Grinding	Grinding the upper and lower teeth resulting in considerable friction and an audible sound (Barnoy, Najdowski, Tarbox, Wilke, & Nollet, 2009; Ford, 1999).
Face-slapping	Forcefully striking one's own the face with an open hand (Healey et al., 2001; Iwata, Dorsey, et al., 1994).
Neck-choking	Forcefully placing hands around the neck in a choking fashion (Iwata, Dorsey, et al., 1994).

Untreated, challenging behaviours such as SIB can severely impact on school and social performance of children with LFA (Glasson et al., 2008; Matson, Sipes, et al., 2011; McClintock et al., 2003). To begin with, a working knowledge of the causes of SIB is crucial to the creation and implementation of effective treatment strategies.

2.5.1 SIB: Possible causes

Historically, the possible causes of SIB have been the topic of numerous investigations and reports. From a psychoanalytic perspective, Kafka (1969) suggested that the sensation of warm blood emanating from SIB wounds soothed people as it was similar to the warmth of a mother's comfort. Pao (1969) suggested that SIB enabled dissociation. Others have suggested that SIB helps people escape demands or is an expression of guilt associated with childhood sexual abuse (Podvoll, 1969; Stone, 1987). Chemically, the pain associated with SIB has been reported to release endorphins which can perpetuate the behaviour (Willer, Dehen, & Cambier, 1981; Winchel & Stanley, 1991).

Koenig et al. (2016) conducted a meta-analysis to investigate if SIB resulted in an absence of pain for people with borderline personality disorder (BPD). A random-effects modelling analysis assessed 32 BPD studies published between 1992 and 2015 based on participants ranging in age from 15.1 to 37.2 years. A Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) analysis focussed on three SIB factors. *Pain threshold*, defined as the time between SIB and the onset of pain. *Pain tolerance*, described as the maximum amount of pain endured. *Pain intensity*, determined by participant numeric and visual analogue ratings. A Hedges' *g* effect size analysis (Hedges & Olkin, 1985) revealed that SIB was associated with a higher pain threshold ($g = 0.76$; high effect), pain tolerance ($g = 0.47$; moderate effect) and lower pain intensity ($g = -0.68$; moderate to high effect) compared to controls.

Nock (2010, p. 352) published a theoretical paper suggesting six "key testable hypotheses" as potential risk factors associated with the occurrence of SIB. His *social learning hypothesis* predicted that SIB may be the result of repeating previously models behavioural observations. A *self-punishment hypothesis* posited that SIB may be a method of self-punishment for perceived wrong-doings. The *implicit attitude/identification hypothesis* suggested that SIB may be a behaviour that is more congruent with one's own self than less injurious behaviour. The *social signalling hypothesis* suggested that SIB may be seen as a method of social communication. The *pragmatic hypothesis* predicted that SIB occurs when it is perceived to be quicker,

more effective and more pragmatic for controlling uncomfortable emotional states or environmental situations. Lastly, the *pain analgesia/opiate hypothesis* suggested that people with lower pain aversions and who are less affronted by the unpleasantness of SIB are more likely to engage. Nock also suggested that the pain associated with SIB may create a euphoric state resulting from higher endorphin secretion and that this may perpetuate further SIB. Similarly, Bresin and Gordon (2013) suggested that humans become more sensitive to opioid based neuropeptides known as beta-endorphins and enkephalins as a result of SIB and that this may perpetuate SIB. Nock (2010) was clear to report that his SIB hypotheses had not yet been empirically supported and that further research was required to determine their validity. Despite the above-mentioned research not being specifically related to people with ASD, it is possibly relevant.

Regarding ASD, the possible causes of SIB can include, but are not limited to, illness, changes in daily routine, environmental changes, sensory dislikes and ineffective communication (Adler et al., 2015). In addition, environments that limit the natural expression of ASD restricted routines, and ritualistic and repetitive movements can cause SIB (Boyd et al., 2013; Green et al., 2006; Lai et al., 2014). SIB exhibited by people with multiple disabilities such as LFA has been associated with impulsive and hyperactive tendencies, anxiousness and/or inadequate sensory regulation (Bright, Bittick, & Fleeman, 1981; Lai et al., 2014). Moreover, and consistent with the heterogeneity and comorbid nature of ASD, SIB is often the result of multiple factors specific to each individual (Adler et al., 2015).

A seminal method of assessing the causes of SIB was described in the *4-dimensional contingency model* (Iwata, et al., 1982; 1994). This model described how the occurrence of SIB can be contingent on social, alone, academic and unstructured play dimensions within the environment. The *social dimension* described as SIB being contingent on social attention. The *alone dimension* explained how SIB can be contingent on the absence of environmental stimulation. The *academic dimension* defined how SIB can be contingent on avoiding academically challenging tasks. Lastly, the *unstructured play dimension* described how SIB can be less likely to occur in environments which contained few demands with unstructured play, close proximity to assistance and praise. This dimensional model is known as the "standard" for analysing the possible causes of SIB and has led to past and present intervention efforts and successes (Beavers, Iwata, & Lerman, 2013, p. 13).

2.5.2 SIB: Possible treatments

Treating SIB exhibited by those with an ID is a complex and challenging task (Smith et al., 2005). This is often made more complex by the numerous SIB types, non-compliance of treatment recipients and pre-existing behavioural and diagnostic comorbidities (Briere & Gil, 1998; Novak, 2003; Richman, 2008). As a result, empirically-based behavioural and pharmacological treatments designed to treat repetitive behaviours such as SIB exhibited by children with ASD are presently lacking in number and efficacy (Boyd et al., 2013). Moreover, treating SIB exhibited by children with ASD before they develop the physical size and strength to injure themselves is of critical importance (Machalicek et al., 2007).

To administer the most effective interventions for people with ASD, it is important to review all of the relevant evidence-based treatment options (Canitano & Scandurra, 2011). Pharmacological treatments, applied behaviour analysis (ABA) techniques, and a combination of the two approaches have been reported to be effective in treating SIB exhibited by children with ASD (Campbell, 2003; Matson, Sipes, et al., 2011; Matson et al., 2012; Neidert, Dozier, Iwata, & Hafen, 2010; Wachtel et al., 2009; Weeden, Ehrhardt, & Poling, 2009). The following sections review the relevant literature regarding these approaches.

2.5.2.1 SIB: ABA behavioural techniques for children with LFA

ABA techniques have been used to achieve positive behavioural outcomes for people with ASD for over 40 years (Hastings & Noone, 2005; Matson & LoVullo, 2008; Matson et al., 2012). ABA was originally implemented to improve communication and reduce severe behavioural difficulties such as SIB in the 1960's by the seminal author and clinician O. Ivar Lovaas (Smith & Eikeseth, 2011). Lovaas (1982) used ABA techniques such as extinction, alternate sensory tasks, overcorrection and timeout for the treatment of SIB. The focus of ABA was at this time, and still is, to increase appropriate and reduce inappropriate behaviours (Koegel, Koegel, & McNerney, 2001). ABA techniques such as a Differential Reinforcement of other behaviour (DRO), differential reinforcement of alternative behaviour (DRA), negative reinforcement, time-out, positive reinforcement, environmental modification and sensory extinction have been reported as effective in the treatment of SIB (Campbell, 2003; Cowdery, Iwata, & Pace, 1990; Durand & Carr, 1987; Harris, Handleman, & Fong, 1987; Hersen, & Sturmey, 2012; Linscheid, Iwata, Ricketts, Williams, & Griffin,

1990; Maag, Wolchik, Rutherford, & Parks, 1986; Matson & LoVullo, 2008; Tiger, Fisher, & Bouxsein, 2009).

Prior to the commencement of ABA, the completion of a Functional Analysis (FA) is recommended as a method of determining if the behaviour is functionally motivated by attention seeking, obtaining a desired object, task avoidance or to obtaining a reinforcement (Hastings & Noone, 2005; Posey et al., 2008). A comprehensive functional assessment, the FA quantitatively assesses the occurrence, non-occurrence of SIB via direct observation (Kahng et al., 2002). To date, FA has been reported in over one thousand published research projects and is mandated for inclusion in educational planning for students with special needs in the USA (Davis, 2015; Iwata et al., 1982; Iwata, Dorsey, et al., 1994). FA methods can detect environmental contingencies associated with the expression of SIB and meaningfully inform treatment approaches (Beavers et al., 2013; Iwata, Pace, et al., 1994).

The function of SIB can be associated with positive social attention, social avoidance or more automated operant functions such as sensory feedback (Barrera, Violo, & Graver, 2007; Iwata et al., 1994). For example, Iwata and colleagues (1994) conducted a seminal epidemiological analysis of SIB based on observational records of 152 people with ID living in hospital inpatient/supported accommodation settings over an 11-year period to determine possible environmental precipitants of SIB. SIB data was derived from 10-second interval recordings throughout 15-minute experimental and control conditions to determine if SIB was reinforced by positive social attention, social avoidance or automated operant functions. Positive social attention and social avoidance functions were found to be the causes of SIB in two thirds of participants. An automated operant punishment function caused SIB in one quarter of the participants. The comprehensive functional assessment used in this research reportedly contributed significantly to the validity of results (Iwata et al. 1994).

Vollmer et al. (1993) used ABA techniques of DRO and non-contingent reinforcement (NCR) to assess if they could reduce SIB such as head hitting, head banging, body hitting, and hand mouthing amongst three adult females with developmental disabilities living in supported accommodation in the USA. A FA prior to the treatment revealed that the participants' SIB functioned as a positive social attention reinforcement. As a result, during the DRO treatment, positive social attention was provided only in the absence of SIB. In addition, during the NCR

treatment, positive social attention was provided at fixed intervals regardless of whether the SIB was occurring. The different treatments were administered between two and four times per day for a duration of 10 to 15 minutes over five consecutive days. SIB occurrences were rated per minute. Vollmer et al. concluded that both DRO and NCR were effective ABA techniques for reducing SIB. Similar to the aforementioned Iwata et al. (1994) epidemiological analysis, the authors attributed their finding to the inclusion of a prior FA.

Rhine and Tarbox (2009) conducted a multielement, single-subject study which used an ABA Functional Communication Training (FCT) technique to reduce severe food rumination SIB exhibited by a six-year-old boy with ASD and suspected ID. The authors explained that FCT was designed to teach alternate behaviours that elicit the same reinforcement as the previously challenging behaviour (Carr & Durand, 1985). Prior to this study, the boy's food rumination had resulted in the decay and subsequent removal of teeth. His rumination was assessed both when he was taught to chew gum as an alternate behaviour compared to when he was not. The treatment order was randomised via a coin toss to avert order effects. The chewing-gum treatment involved the author pairing the presentation of a piece of chewing-gum with a verbal prompt which was non-contingent on food rumination. If the boy did not accept the chewing-gum, it was held 30cm from him and he was continuously prompted at 3 second intervals until it was accepted. The author did not intervene during the non-chewing gum periods. The authors reported that the boy's rumination was less frequent during the chewing gum treatment phase when compared to the non-chewing gum treatment phase. This result was maintained at 1, 2 and 3-month follow-up.

Barnoy et al. (2009) reported the results of a single-case reversal BABCB design which compared the use of ABA several methods designed to reduce the frequency of teeth-grinding SIB for a six-year-old girl with ASD and suspected ID. It was hypothesised that that intervention condition would reduce SIB more than the non-intervention condition. Contingent on her exhibiting teeth-grinding SIB, the authors administered one of three treatments. Firstly, when she began to grind her teeth, the authors verbally prompted her to "Say Ah" whilst physically press her chin downward (Barnoy et al., 2009, p. 846). Alternately, in response to the teeth-grinding SIB, the authors ignored the behaviour and did not intervene. During the third treatment, the authors provided a verbal prompt only in response to teeth-grinding SIB. The authors found that a combination of verbal prompting and physical intervention reduced SIB.

Despite the high quantity of positive ABA results reported in the literature, according to Lai et al. (2014), ABA research could benefit from additional empirical support in relation to the treatment of aggressive behaviours such as SIB. Further, it has been reported that ABA techniques can be time, cost, process and training intensive (Hastings & Noone, 2005; Matson, Sipes, et al., 2011; Posey et al., 2008). In addition, conducting a FA prior to ABA, it requires specially trained, skilled and experienced analysts (Hastings & Noone, 2005). Therefore, it is pertinent to consider the established and growing body of knowledge regarding the efficacy of pharmacology therapies for the treatment of SIB.

2.5.2.2 SIB: Pharmacological therapies for children with LFA

Contemporary literature reports that approximately one quarter of children with ASD aged three to 14 years in the US are undergoing pharmacological treatment (Mohiuddin & Ghaziuddin, 2013). Pharmacological therapies act to biologically alter brain function as a method of managing difficult behaviours (Westen, Burton, & Kowalski, 2006). These types of treatments are commonly used to manage challenging behaviour such as SIB displayed by people with ASD when behavioural treatments in alone have proven ineffective (Posey et al., 2008). When successful, pharmacological therapies can result in therapeutic compliance and greater school participation for children with ASD and SIB (Politte & McDougle, 2014). In extreme life threatening situations, pharmacological interventions can act as an "emergency restraint" to treat SIB (Matson & LoVullo, 2008, p. 71). As such, it is not uncommon for primary carers of children with ASD to consider administering pharmacological therapies (Williamson & Martin, 2012). Despite this, the efficacy of these treatments can vary from individual to individual and side–effects can be challenging (Brandes, 2009; Matson, Sipes, et al., 2011).

Typical and atypical antipsychotic medications are classified under the psychotropic medication class of pharmacological therapy (Williamson & Martin, 2012). Of these, atypical antipsychotics have been reported to be the most effective in treating irritability inclusive of SIB amongst children with ASD (Benvenuto, Battan, Porfirio, & Curatolo, 2013; Politte & McDougle, 2014). To date, the USA Food and Drug Administration (FDA) has approved the use of two specific atypical antipsychotic medications for the pharmacological treatment of irritability exhibited by children with ASD: risperidone and aripiprazole (Politte & McDougle, 2014; U.S. Food and Drug Administration, 2006; Williamson & Martin, 2012).

Risperidone was approved by the FDA in 2006 for the treatment of irritability amongst children with ASD aged 5 to 16 years (Williamson & Martin, 2012). This form of pharmacological therapy blocks reuptake receptors to the neurotransmitters for dopamine D_2 and serotonin 5-HT_{2A} (Rosenbaum, Hyman, Labbate, & Fava, 2005). To date, risperidone is the most researched and commonly applied pharmacological therapy for reducing aggressive behaviour such as SIB amongst children with ASD (Lai et al., 2014; Mohiuddin & Ghaziuddin, 2013; Politte & McDougle, 2014; Stigler & McDougle, 2008). This has eventuated as a result of the following two large-scale, double-blind, randomised and placebo-controlled trials.

McCracken et al. (2002) reported the results of an 8-week risperidone randomised, double-blind and control versus placebo pharmacological therapy trial. The authors collected Aberrant Behavior Checklist (ABC; Aman & Singh, 1986) and Clinical Global Impressions – Improvement (CGI-I) scale data from 101 children aged five to 17 years with ASD and irritability including SIB at multiple research venues in the USA. The ABC (Aman & Singh, 1986) included an irritability subscale of 15-items which determined temper outbursts, aggressive tendencies, mood labilities and SIB (Marcus et al., 2011). Of the 101 participants, 49 were administered risperidone and 52 a placebo. The authors reported that a daily dose of 1.8 mg plus or minus 0.7mg of risperidone resulted in a 56.9 percent reduction in irritability within the risperidone group as opposed to 14.1 percent in the placebo group. However, adverse effects such as weight gain, saliva drooling, fatigue, sleepiness and dizziness were also reported (McCracken et al., 2002).

Employing a similar randomised, double-blinded, control versus placebo trial Shea et al. (2004) investigated the effects of risperidone on 79 children aged five to 12 years with ASD over an 8-week period. The outcome measures collected by the authors were the ABC, Clinical Global Impressions – Change (CGI-C) scale and Nisonger Child Behavior Rating Form (Tassé, Aman, Hammer, & Rojahn, 1996). Children who were administered risperidone recorded significantly lower irritability scores and an overall improvement in behaviour compared to the placebo group. However, similar to McCracken et al. (2002), risperidone caused weight gain, as well as systolic blood pressure and heart rate elevations (Shea et al., 2004).

Risperidone can cause hyper-salivation, sedation, the development of random and involuntary movements known as tardive dyskinesia and excessive prolactin secretion known as hyperprolactinemia (International Society for the Study of Trauma

and Dissociation, 2011; Posey et al., 2008). Furthermore, this form of pharmacological therapy has not yet achieved unequivocal scientific endorsement and can be used for long periods of time without standardised dosage parameters or administration schedules for people with ASD (Canitano & Scandurra, 2011). In sum, the favourable outcomes of risperidone appear to be limited by its numerous adverse effects.

Aripiprazole was approved in 2009 by the FDA for use amongst children with ASD aged 6 to 17 years for the treatment of irritability inclusive of SIB (Politte & McDougle, 2014). This form of pharmacological therapy acts to partially block neurotransmitter reuptake of dopamine D_2 and serotonin HT_{1A} receptors whilst also blocking the uptake of serotonin 5-HT_{2A} (Erickson, Stigler, Posey, & McDougle, 2010). Similar to risperidone, aripiprazole has been reported to be effective in the treatment of SIB exhibited by those with ASD (Benvenuto et al., 2013; Lai et al., 2014). Two recent studies of note indicate the possible efficacy of taking aripiprazole as a pharmacological therapy for the reduction of irritability for ASD.

Owen et al. (2009) employed a randomised, multi-centre, double-blind, placebo versus aripiprazole pharmacological therapy trial amongst 98 children with ASD aged six to 17 years. The aripiprazole group were administered doses of 5 mg, 10 mg, or 15 mg each day over 8 consecutive weeks. The authors used the ABC irritability subscale and the CGI-I scale to assess irritability. The aripiprazole group recorded significantly lower irritability scores on both the ABC and CGI-I. Despite this result, aripiprazole caused "sleepiness, vomiting, fatigue, increased appetite, and tremors" (Owen et al., 2009, p. 1538).

Marcus et al. (2011) investigated the effects of daily aripiprazole administered for 52 consecutive weeks on 199 people with ASD aged 6 to 17 years across 53 research sites within the USA. Participants were assessed on the ABC irritability subscale and the CGI-I scale. Aripiprazole was found to significantly reduce irritability over the 52-week trial. However, the adverse effects experienced by the participants included an increased appetite, weight gain, sleeplessness and vomiting.

To assess the efficacy of both risperidone and aripiprazole pharmacological therapies, Politte and McDougle (2014) conducted a review of literature published from 1999 to 2012 including participants with pervasive developmental disorders such as ASD. Risperidone was found to be well tolerated for the treatment of irritability such as SIB, however its adverse effects such as gaining weight, increasing prolactin levels and drowsiness were limiting. Aripiprazole, was also found to be well tolerated

for the treatment of irritability without increasing levels of prolactin. Despite the fewer adverse effect of taking aripiprazole, the small number of publications presents a substantial limitation. In sum, while risperidone and aripiprazole are endorsed by the FDA for the treatment of irritability including SIB amongst children with ASD, the adverse effects of both are limiting (Politte & McDougle, 2014; Williamson & Martin, 2012). Risperidone and aripiprazole are also listed on The Pharmaceutical Benefits Scheme in Australia (Australian Government Department of Health, 2016).

The heterogeneous nature of ASD indicates that no single behavioural or pharmacological therapy will be efficacious for all children with the diagnosis (Stahmer, 2014). Further, those with ASD who exhibit SIB can often be medication resistant and experience adverse effects (Adler et al., 2015). Therefore, alternative treatment options are needed. Interestingly, people with ASD can participate in music therapy (Green et al., 2006; Papagiannopoulou, 2015).

2.6 Music Therapy

"Music Therapy is a behavioural science concerned with human behaviour and is based on a scientific approach" (Davis, Gfeller, & Thaut, 1992, p. 6). Music Therapy (MT) includes structured or unstructured improvisation, playing music, creating music and music listening (Wheeler et al., 2005). Music therapists aim to enhance, preserve and reinstate the well-being of humans through the use of music (Finnigan & Starr, 2010). Moreover, music therapists can work with people across multiple settings, including children who have profound disabilities such as LFA (Avers, Mathur, & Kamat, 2007).

As an established mode of healthcare, MT has been used longitudinally to treat social, emotional, cognitive and physical difficulties experienced by people of all ages (Finnigan & Starr, 2010). MT can promote health, avert illness and provide a safe and non-invasive therapeutic approach (Hooper et al., 2010; Mra´zova´ & Celec, 2010; Munro & Mount, 1978). Throughout the last century in medicine, music therapists have adopted an integral role in the care and treatment of patients (Stockman, 2008).

A meta-analysis published by Gold et al. (2004) investigated the efficacy of 11 group and individual MT studies published between 1970 and 1988 designed for children and adolescents living in the US, UK, Austria and Germany. An analysis of MT post-test data across the studies revealed a large Cohen's *d* overall mean effect size of $d = .99$. Of interest, large Cohen's *d* effect sizes were also found regarding the

efficacy of MT for children with behavioural disorders (d = .78) and developmental disorders (d = .65). The authors concluded that MT was clinically useful.

Further, a meta-analysis conducted by Pelletier (2004) evaluated the effectiveness of MT on those under and over the age of 18 years. Twenty-two peer-reviewed papers published between 1985 and 2004 were assessed. The studies quantitatively assessed the efficacy of MT applied in medical and university settings designed to reduce stress induced by artificial and naturally occurring life events. Heart rate, behavioural observations and self-reports were recorded as indicators of stress. The author reported an overall mean Cohen's d effect size of d = .6711. It was concluded that music listening coupled with MT, progressive muscle relaxation coupled with MT, guided imagery coupled with MT, and verbal suggestion coupled with MT significantly reduced stress amongst participants.

More recently, a meta-analysis conducted by Standley (2012) assessed the potential benefits of MT on the behaviour and development of preterm infants being treated in neonatal intensive care units. Thirty peer-reviewed experimental single-case and group MT research papers published between 1950 and 2010 were assessed. Preterm infant measures such as heart and respiration rate, feeding, body weight, blood pressure, oxygen saturation and behavioural observations were analysed. Infants were exposed to recorded and live music combined with and without human touch. The author reported a large overall mean Cohen's d effect size of d = .82. MT was reported to reduce the length of neonatal intensive care unit stay, improve oxygen levels, feeding, sleep and pacifying.

Most recently, Chang et al. (2015) published a meta-analysis of ten peer reviewed, randomised controlled trials (RCTs) using a PRISMA framework. Effect sizes of quantitative MT studies including older adults with dementia published from 2000 to 2014 were analysed. Challenging behaviours were measured via the Neuropsychiatric Inventory (NPI), the complete, short form and Chinese version of the Cohen–Mansfield Agitation Inventory (CMAI) along with the Behavioural Pathology in Alzheimer's disease instrument. Anxiety was assessed via the Hamilton Anxiety Rating Scale, NPI and Rating Anxiety in Dementia (RAID). Depressive mood was measured by the Multidimensional Observation Scale for Elderly Subjects, Geriatric Depression Scale, NPI and the Cornell Scale for Depression in Dementia. Cognitive abilities were measured using the Mini-Mental Status Examination of older adults. A Hedges' g (Hedges & Olkin, 1985) effect size analysis revealed that MT

reduced challenging behaviours with moderate to high effect (g = -0.66), anxiety with moderate effect (g = -0.51), depressive mood with moderate effect (g = -0.39) and improved cognitive abilities with a small effect (g = 0.19). The studies included in this meta-analysis included both passive and active MT. As such, both passive and active MT have the potential to elicit positive effects (Ockelford, 2013 as cited in Fancourt, Ockelford, & Belai, 2014).

2.6.1 Active music therapy

Active MT can range from educational music lessons and workshops to using music therapeutically to reflect emotions and patterns of thinking (Fancourt et al., 2014). Finnigan and Starr (2010) conducted a study in which it was hypothesised that active MT would increase social responsiveness via eye contact, imitation and turn-taking frequency counts and decrease social avoidance via gaze avoidance, rejection of toys, rejecting and moving away from adults' frequency counts. These measures were applied to a three-year-old girl with ASD and a suspected ID. The researchers used an ABCD single-subject alternating treatment design where the behaviour of the child was observed, recorded and coded across baseline, treatment, method of effective treatment return and follow-up phases. Social response and social avoidance behaviours were observed as the child interacted with the music therapist during 12 active MT sessions, four times per week over 29 weeks at the child's school and home. The authors reported that active MT resulted in significant increases in social response and reductions in social avoidance. Specifically, singing songs with the young girl was found to be most effective in increasing her social responsiveness.

Pasiali, LaGasse, and Penn (2014) conducted a single group, pre-test and post-test active MT intervention for nine people in a school setting aged 13 to 20 years with neurodevelopmental disorders including ASD. MT was administered over a 6-week period inclusive of eight sessions which were 45 minutes in length. Subjects participated in active MT tasks such as "…structured drumming/rhythm experiences, and structured/unstructured improvisation… singing songs and chants, using body percussion…" which formed the musical attention control training treatment (Pasiali et al., 2014, p. 344). Attention skills were measured via the Test of Everyday Attention for Children (TEA-Ch; Manly et al., 1999) to determine the feasibility of the MT protocol. Active MT resulted in improved selective attention and attention control/switching skills. However, history effects such as breaks in the school year between TEA-Ch administrations may have confounded the results.

2.6.2 Passive music therapy: Music listening

Passive MT participation, operationally defined here as *music listening*, involves exposure to pre-recorded or live music (Ockelford, 2013 as cited in Fancourt et al., 2014). Music listening has been linked to favourable treatment outcomes for people with neurodevelopmental disorders such as ASD (Hooper et al., 2010).

Using a single-case ABAB withdrawal design, Kern, Wolery, and Aldridge (2007) investigated the effects of routine-based songs sung to two boys aged 3 years with ASD by their child care teachers as part of a morning greeting routine. Each song was composed specifically for each boy by a music therapist, in consultation with a teacher and primary carer, to correspond with the tasks require of the school morning routine. Before the research began, it was determined that both boys experienced severe difficulties during the morning greeting routine at school as indicated by school refusal with oppositional and defiant behaviour. The composition included typical classroom activities such as entering the classroom, putting personal belongings into pigeon holes, greeting teachers and classmates, farewelling primary carers, and participating in free play. During the A conditions of the ABAB design, the boys attempted their morning routine without the song whereas during the B conditions the song was sung by the classroom teacher as the boys attempted the morning routine. Direct observations of the morning greeting routine, task compliance, adult physical/verbal routine prompting and non-prompting were recorded by a triad of raters including a trained music therapist, education assistant and research assistant. The authors concluded that music listening helped both boys complete their morning routine with greater compliance and more positive responses in the areas of independence, teacher and peer greetings, participation and social play. Furthermore, the boys' classmates made an increased number of social approaches. However, the musical treatment was dependent on having access to a qualified music therapist. Further, a FA was not conducted to determine the primary factor associated with the severe difficulties during the morning greeting routine. As a result, the findings may have been associated with an extraneous factor other than the music.

Katagiri (2009) analysed the effect of music listening on twelve school children with ASD in Japan aged between 9 and 15 years (M = 11. 5 years). The authors used a pre-test and post-test counterbalanced design to administer four conditions: not teaching a selected emotion, teaching a selected emotion verbally, teaching a selected emotion verbally whilst listening to improvised piano music which matched the

emotion, and teaching a selected emotion through the singing of songs about the emotion. Each condition included the teaching of emotions of happiness, sadness, anger and fear, and was administered eight times every 2 weeks for a duration of 30 minutes. The pre-test and post-test instruments assessed the participants' understanding of emotions via facial expressions and situations as indicators of emotion encoding and decoding abilities. Of the four conditions, those who learned an emotion via verbal instruction accompanied by music listening improved their understanding of the selected emotion significantly more than others. The internal validity of this study was limited by the absence of inter-rater reliability data.

Hooper et al. (2010) analysed the arousal levels of 96 adults aged between 18 and 69 years both with and without a mild ID (FSIQ 55 to 77 points) after listening to a mixture of calming and stimulating instrumental musical segments. The authors assessed arousal based on participant ratings of the 24-item UWST Mood Adjective Checklist (UMACL) as developed by Matthews, Jones, and Chamberlain (1990). The group without an ID listened to musical segments lengths of 2 minutes and 30 seconds before completing the UMACL in their homes. Whereas the group with an ID listened to musical segments lengths of 30 seconds, to account for their ID, in a controlled environment. The authors found no significant differences in levels of arousal recorded between groups after listening to the instrumental segment entitled "*Yesterday* (12 cellists)" from the CD "Beatles in Classics The 12 Cellists of the Berlin Philharmonic" (Hooper et al., 2010, p. 25). That is, people with an ID performed similarly to those without an ID. At the conclusion of their paper, the authors suggested that the underlying cause of challenging behaviour for those with an ID may be unregulated arousal. Further, it was hypothesised that music listening may be suitable for reducing arousal among this population as it requires the ability to listen to sounds only.

Hooper, Carson, and Lindsay (2012) conducted a study in which they assessed the potential of music listening to reduce anxiety and disruptive lunchtime behaviours such as physical and verbal aggression and SIB amongst a group of 30 adults with varying levels of ID. Behaviour was recorded as participants ate lunch in their homes or in the dining rooms of respite facilities. On the first day of the two-day treatment, participants were randomly allocated to music listening or no music listening conditions. Those allocated to the music listening condition listened to calming instrumental music played through earphones plugged into an MP3 player which hung from a lanyard around the neck of the participant. The following day, the music

listening and no music listening conditions were reversed. Five of the 30 participants with the most severe disabilities withdrew because they refused to wear the MP3 player and earphones. Of interest, listening to calming music reduced self-harm, verbal repetitiveness, and restlessness at mealtimes for three of the participants.

Although music listening has been reported to positively influence behaviour of those with ASD, it would be premature to suggest that all people with LFA will react similarly. However, music which acts to regulate an internal process such as arousal may also minimise maladaptive and externalised behaviours such as SIB for those with limited communicative abilities, as is often the case for people with LFA (Ford, 1999). Moreover, people with LFA may benefit from MT as an augmentative approach to existing therapies which are designed to reduce stress (Avers et al., 2007; Boso, Emanuele, Minazzi, Abbamonte, & Politi, 2007; Mra´zova´ & Celec, 2010).

2.7 Arousal

Arousal is a "state of alertness, from being asleep to being in a highly energised state" (Dibben & Williamson, 2007, p. 576). Humans regulate their levels of arousal by releasing biological agents tasked with restoring the body from an over-aroused or under-aroused state back to a steady state; a process known as homeostasis. Measuring and analysing these biological agents, otherwise known as biomarkers of arousal, can provide an objective measure of arousal; and when detected in saliva, can provide a relatively non-invasive and socially appropriate method of determining the levels of arousal amongst specific cohorts such as those with LFA (Fortunato, Dribin, Granger, & Buss, 2008). However, it is important to consider that biological responses to arousal can vary from human to human as people perceive and process arousing life-events differently (Bauer, Quas, & Boyce, 2002; Hirvikoski & Blomqvist, 2014). Cortisol and alpha-amylase are biomarkers of arousal and are components of the hypothalamic-pituitary-adrenal axis and autonomic nervous system respectively.

2.7.1 The hypothalamic-pituitary-adrenal axis and cortisol

A primary function of the hypothalamic-pituitary-adrenal (HPA) axis is to help the body adapt to internal and external stress by releasing the steroid hormone cortisol (Kandel, Schwartz, Jessell, & Schwartz, 2000; Koss et al., 2014; Taylor et al., 2013; Zinke, Fries, Kliegel, Kirschbaum, & Dettenborn, 2010). As illustrated in Figure 2.1, in response to psychological or physiochemical stressors, corticotrophin-releasing hormone (CRH) is secreted by the hypothalamus. CRH in turn stimulates secretion of adrenocorticotropic hormone (ACTH) by the anterior pituitary which in turn elicits the

release of cortisol by the adrenal cortex into the circulation, raising the concentration of cortisol in blood and in other body fluids, including saliva and urine (Bauer et al., 2002; Bitsika, Sharpley, Andronicos, & Agnew, 2015; Fortunato et al., 2008; Rudolph, Troop-Gordon, & Granger, 2010). CRH and ACTH secretion can be inhibited by rising cortisol concentration as part of a feedback loop (see Figure 2.1). This negative feedback effect can be modulated in response to CNS triggers which result in higher than usual cortisol concentrations. However, a low circulating cortisol concentration will also increase CRH and ACTH secretion due to a lack of negative feedback.

The circadian rhythm of cortisol defines how the concentration of the hormone decreases during the day. It is lowest during the first half of the night, rises during the second half of the night and peaks in the morning (Fries, Dettenborn, & Kirschbaum, 2009). Cortisol detected within saliva is known as salivary cortisol (sCort) and can be measured in micrograms per decilitre (μg/dL) of biologically active cortisol concentration in saliva samples which correlate with free cortisol concentrations in plasma (Kirschbaum & Hellhammer, 1989).

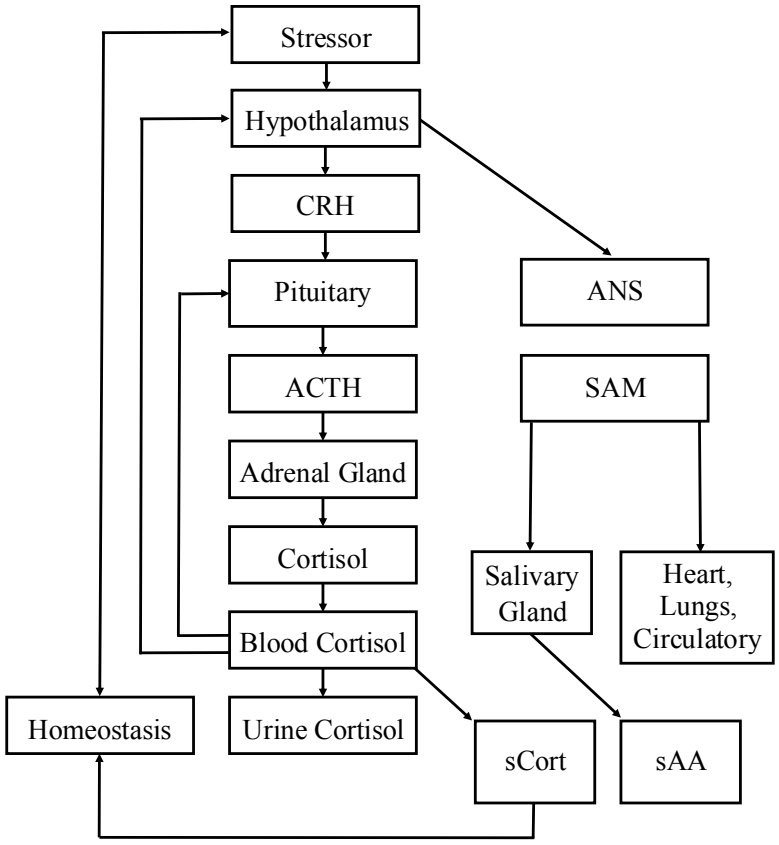


Figure 2.1 The human stress response; CRH: corticotrophin-releasing hormone; ANS: autonomic nervous system; SAM: sympathetic adrenal medullary system; ACTH: adrenocorticotropic hormone; sCort: salivary cortisol; sAA: salivary alpha-amylase.

Cortisol helps to sustain numerous physiological processes in the face of stress (Bitsika et al., 2015; Sapolsky, Romero, & Munck, 2000). For example, cortisol functions to increase blood glucose concentration, an important source of energy for cells – this is one of numerous metabolic, cardiovascular and neurologic effects enabling individuals to cope with acute or enduring and stressful events (White & Mulligan, 2009). The HPA axis raises cortisol activity, therein biological arousal levels, to meet the demands of stressors (Bitsika et al., 2015). Alternately, low cortisol concentrations can be the result of the HPA axis becoming desensitised to repetitive or extended periods of extremely high biological arousal (Bitsika, Sharpley, Sweeney, & McFarlane, 2014; Dienstbier, 1989).

Bitsika et al. (2015) assessed two measures of anxiety amongst 150 Australian boys with ASD, an average age of 11.1 years and a FSIQ of over 70 points. Firstly, saliva samples were collected 30 minutes after waking then between 2 pm and 4 pm. The samples were analysed for sCort concentrations to determine biological anxiety levels. Secondly, anxiety symptoms were rated by the boys and their primary carers using 8 generalised anxiety disorder (GAD) items from the fourth revision of the Child and Adolescent Symptom Inventory (CASI-4). The sCort concentrations and CASI-4 ratings were analysed individually and jointly to determine possible correlations. The authors found that average sCort concentrations reduced consistently from morning to afternoon. Further, the frequency of CASI-4 anxiety symptoms was higher amongst the boys with ASD when compared to existing CASI-4 norms. Despite some statistical inconsistencies within the sample, sCort was reported to be a valid biological measure of anxiety among boys with ASD who also recorded a consistent reduction from morning to afternoon.

For those with ASD, environmental demands and aversions can significantly influence biological levels of arousal (Kidd et al., 2012). For example, children with ASD can become hyper-responsive to acute periods of environmental arousal, which is often evident in school contexts (Richdale & Prior, 1992; Tordjman et al., 1997). Periods of cortisol overproduction "might also be a factor in exacerbating the already present difficulties associated with the behavioural manifestations of ASD" such as challenging behaviours (Bitsika et al., 2015, p. 201). Further, when arousal remains heightened for extended periods, it can cause physiological damage to the body of a child (Avers et al., 2007). As such the HPA axis and subsequent cortisol concentration variations recorded among children with ASD can produce inconsistent results

(Corbett, Mendoza, Abdullah, Wegelin, & Levine, 2006; Corbett, Mendoza, Wegelin, Carmean, & Levine, 2008; Corbett, Schupp, & Lanni, 2012; Tordjman et al., 2014; Zinke et al., 2010). In light of these inconsistencies, additional biomarkers of arousal may be helpful in determining treatment effect. Just as the HPA axis functions to regulate arousal, the autonomic nervous system (ANS) also responds to arousing events which disturb periods of homeostasis (Kidd et al., 2012).

2.7.2 The autonomic nervous system and alpha-amylase

In addition to other biological responses which affect the cardiovascular, metabolic and digestive systems, the ANS, otherwise known as the "fight or flight" system, helps the body survive when challenged (Koss et al., 2014, p. 837). As illustrated in Figure 2.1, in response to a stressor, increased amounts of alpha-amylase are released via activation of the sympathetic adrenal medullary (SAM) axis of the ANS (White & Mulligan, 2009). As the SAM is an axis of the ANS, the term ANS is herein used operationally. Activation of the ANS stimulates endocrine, exocrine and life-preserving reactions from vital organs such as the pancreas, heart, lungs and circulatory system (Bitsika et al., 2014). One of the products of this process is an increased release of the metabolising enzyme named alpha-amylase into saliva (Granger et al., 2006; Kandel et al., 2000).

Alpha-amylase is present in numerous tissues; pancreatic amylase acts to break down dietary starch into disaccharides and trisaccharides in the small intestine. These smaller saccharides are then converted into glucose to provide energy for "fight or flight" purposes (Gurung, Ray, Bose, & Rai, 2013). Starch is often metabolised in the oral cavity, therefore, alpha-amylase is detectable in saliva as a biomarker of arousal (Gurung et al., 2013; Zakowski & Bruns, 1985).

There is a salivary isoform located within salivary glands known as salivary alpha-amylase (sAA) (Fortunato et al., 2008; Granger, Kivlighan, El-Sheikh, Gordis, & Stroud, 2007; Saarikallio & Erkkilä, 2007). The ANS increases sAA activity by both elevating secretion of the enzyme into saliva and reducing the amount of total saliva production (White & Mulligan, 2009). As such, sAA activity is both a valid biomarker of ANS activity and an overall indicator of the body's response to immediate and ongoing arousal (Koss et al., 2014; Nater & Rohleder, 2009; Nigg, 2006; Taylor et al., 2013; Wetherell et al., 2006). The sAA enzyme can be measured in units per millilitre of alpha-amylase activity in saliva samples (U/mL), and is predictive of the secretion

of a neurotransmitter also tasked with arousal management in plasma called norepinephrine (Chatterton, Vogelsong, Lu, Ellman, & Hudgens, 1996).

Even though the HPA axis and ANS are separate systems, they share responsibility for regulating arousal and returning the body to a state of homeostasis (Bauer et al., 2002; Chrousos & Gold, 1992; Rudolph et al., 2010). However, the HPA axis passively reacts to unforeseeable stressors whereas the ANS actively defends the body against foreseeable stressors (Henry, 1993; Lundberg & Frankenhaeuser, 1980; Schachter & Singer, 1962). For example, the HPA axis passively reacts to situations requiring evasive action whereas the ANS actively defends/prepares the body for stressful upcoming events. Of interest, higher sCort concentrations, as indications of HPA axis activity, have been linked to challenging behaviours such as SIB exhibited by adults with developmental disabilities (Symons, Sutton, Walker, & Bodfish, 2003).

2.7.3 sCort and sAA

Despite sCort and sAA providing biological measures of arousal, there are numerous differences between the biomarkers. Within saliva samples, changes in sAA activity are detectable more immediately than those of sCort (Quas, 2011). The HPA axis is slower to react to demand conditions than the ANS because sCort concentration elevation requires activation of the hypothalamus, pituitary and adrenal structures and hormonal synthesis whereas sAA is the result of neural stimulation of salivary glands and the release of pre-formed amylase granules (Bitsika et al., 2014). As a result, on average, sAA activity and sCort concentration peaks approximately 10 and 20 minutes, after being exposed to a demand condition respectively (White & Mulligan, 2009). Further, poor sleep can be associated with higher sCort concentrations among people with ASD (Chicoine et al., 2013). In addition, sAA activity and sCort concentrations can vary depending on the time-point of saliva collection. Opposite to sCort concentration peaking in the morning and being lowest in the evening, sAA activity is lowest in the morning and peak in the evening (Granger et al., 2006).

Collecting saliva samples for the analysis of biological markers of arousal is a non-invasive and socially appropriate method of measuring HPA axis and ANS arousal regulation amongst individuals with special needs on a case by case basis (Davis & Granger, 2009; Fortunato et al., 2008; Gordis et al., 2006; Taylor et al., 2013). Further, the validity of sAA activity and sCort concentration data can be bolstered by standardising saliva sample collection time-points, assay protocols and saliva sample storage (White & Mulligan, 2009). Overall, the inclusion of biomarkers into applied

research can result in a greater understanding of how behaviour, physiology and psychosocial factors can impact on behaviour change (White & Mulligan, 2009).

2.7.4 The HPA Axis and ANS

Research designed to determine the potential links between arousal regulatory systems such as the HPA axis and ANS can help us understand the possible physiological underpinnings of behaviour (Bauer et al., 2002). Therein, given that the HPA axis and ANS both function to regulate arousal, when studied in combination, subsequent findings may be meaningful to the body of knowledge (White & Mulligan, 2009). Moreover, comprehensive assessments of behavioural and biological data should be gathered across multiple contexts and systems (Allwood et al., 2011; Bauer et al., 2002). Most appropriate for non-typical and externalising behaviour such as SIB exhibited by children, such investigations may reveal potential differences between psychological and biological arousal amongst individuals (Bauer et al., 2002; E. P. Davis & Granger, 2009; Gordis et al., 2006).

Changes in the HPA axis as indicated by changing cortisol activity are best detected when the body is challenged to adapt to demands which maximise arousal (Kudielka & Wüst, 2010). A research-based protocol which has been developed to create such a demand, is known as a trier social stress test (Kirschbaum et al., 1993).

2.8 The Trier Social Stress Test

Research designed to investigate the HPA axis and ANS activity in response to demand conditions among children with ASD has been reported as potentially meaningful (Corbett et al., 2012). Preliminary reports suggest that when arousal levels are symmetrical, children with ASD can present with compliant and functional behaviour (Joosten & Bundy, 2010). Specific to this research, assessing the presence or absence of a symmetrical or asymmetrical association between the HPA axis and ANS may prove seminal to finding out more about the origins of challenging behaviour exhibited by children with ASD such as SIB (Kidd et al., 2012). An empirically validated method of eliciting a demand condition for the assessment of an association between the HPA axis and ANS is known as a Trier Social Stress Test.

The Trier Social Stress Test (TSST) was first published by Kirschbaum, Pirke, and Hellhammer (1993). In this study, 20 male University of Trier students in Germany provided saliva samples before and after being exposed to several demand conditions (Kirschbaum et al., 1993). The participants were required to deliver a five-minute speech on a self-chosen topic to a panel of three confederate employers.

Following this, the same participants were asked to "serially subtract the number 13 from 1,022 as fast and as accurately as possible" (Kirschbaum et al., 1993, p. 77). If participants made calculation errors during this task, they were prompted by a panel member to recommence from the beginning. Of further demand, participants were informed that they were being filmed whilst attempting the tasks for a behavioural analysis and audio was being recorded for a frequency of voice analysis. Based on the analysis of saliva samples collected during the research, the authors concluded that sCort concentration increased significantly as a result of administering the TSST, hence validating the protocol as an effective research-based demand condition.

Since the initial TSST trial, the protocol has been used in numerous studies designed to investigate behavioural and biological responses to demand conditions (Kirschbaum et al., 1993). Khalfa, Bella, Roy, Peretz, and Lupien (2003) published a study which administered the TSST protocol in addition to the collection of saliva samples amongst two groups of adults. Participants allocated to a music listening group listened to "relaxing excerpts from Enya, Vangelis and Yanni … delivered via loudspeakers" immediately after a TSST whereas a no music group listened to silence (Khalfa et al., 2003, p. 375). The music listening group recorded lower sCort concentrations after the TSST compared to those who listened to silence. One year later, Miluk-Kolasa, Obminski, Stupnicki, and Golec (1994) published a similar result for adults who listened to music after being verbally told stressful information about an upcoming surgery.

Moreover, the TSST has been used in behavioural and biological research for those with depression (Burke, Davis, Otte, & Mohr, 2005), anxiety (Roelofs et al., 2009), attention deficit hyperactivity disorder (Lackschewitz, Hüther, & Kröner-Herwig, 2008) and Schizophrenia (Brenner et al., 2009; Kirschbaum et al., 1993). As a result, the TSST has been reported to be a valid and reliable method to facilitate an arousal response in research participants of all ages and genders (Kudielka, Buske-Kirschbaum, Hellhammer, & Kirschbaum, 2004). Further, the TSST has become known for measuring human response to demand conditions via the analysis of saliva samples (Kudielka & Wüst, 2010). Despite this, additional TSST research is needed to further assess its validity across more varied experimental conditions and participant cohorts (Kudielka & Wüst, 2010).

2.9 The Trier Social Stress Test for Children

Buske-Kirschbaum et al. (1997) adapted the TSST to account for social stressors experienced by children. The authors published research using what they named the Trier Social Stress Test – Child version (TSST-C) designed to assess HPA axis activity (Lanni, Schupp, Simon, & Corbett, 2012). Typically developing children aged eight to 14 years were exposed to a 20-minute task including a 5-minute preparation, 5-minute speech to explain how a story finished, 5-minute period of a verbally subtracting increments of 7 from 758 in front of an audience then a 5-minute debriefing. Arousal levels were assessed by analysing saliva samples collected at three time-points before and four time-points after the demand tasks for changes in sCort concentrations. The authors concluded that the TSST-C protocol was effective in eliciting demand conditions for children as it resulted in significant increases in sCort concentration.

The TSST-C has since been validated as research protocol for the assessment of children's biological reactions to a demand conditions via the assessment of sCort concentration and sAA activity as indicators of the HPA axis and ANS (Monteleone et al., 2011). Subsequent publications applied and validated the TSST-C protocol for eliciting demand conditions for research including children (Corbett et al., 2012; Kudielka et al., 2004 & Kirschbaum, 2004). To the researcher's knowledge, the TSST-C has not yet been used in research designed to treat children with LFA. However, the TSST-C protocol as published by Buske-Kirschbaum et al. (1997) would require further adaptation to accommodate for and assess the behaviour of children with LFA.

2.10 The Kagan et al. (1994) and Bauer et al. (2002) Models of Child Behaviour

Kagan et al. (1994) suggested the existence of two physiological child temperament profiles which were thought to be observable in children: inhibited temperament and uninhibited temperament. Children with an inhibited temperament profile were reported to have an over-aroused central nervous system (CNS) which resulted in the expression of internalising behaviours such as withdrawal and avoidance. However, children of an uninhibited temperament were thought to experience an under-aroused CNS which resulted in externalising behaviours such as hyperactivity and aggression. As Kagan et al. (1994, p. 140) stated: "We must show meaningful links between behaviour and physiology". As a result of Kagan's work, research designed to investigate potential links between behaviour and physiology

amongst children was undertaken; such as the additive and interactive models proposed by Bauer, Quas, and Boyce (2002).

Bauer et al. (2002) identified two possible explanations as to how the HPA axis and ANS activity may function to precipitate challenging behaviour such as SIB exhibited by children who are deemed to be at risk of behaviour problems. Bauer et al. (2002) hypothesised that the HPA axis and ANS which function to regulate arousal in children should be assessed in combination as opposed to isolation with the intention of truly understanding the origin of their challenging behaviours. The two models identified by Bauer et al. (2002) were the symmetrical *additive model* and the asymmetrical *interactive model*.

The additive model predicted that when the HPA axis and ANS activity were either both low ("deactivation") or both high ("activation"), the risk of challenging behaviour was elevated (Bauer et al., 2002, p. 107). In sum, the likelihood of challenging behaviour occurring was highest when there was a symmetrical association between the HPA axis and ANS activation (Koss et al., 2014). Conversely, the model predicted that the risk of challenging behaviour expression was least likely when the association between the HPA axis and ANS activity was asymmetrical. Restated, challenging behaviour is least likely to occur when the HPA system is highly activated and the ANS is at a low level of activation and vice versa (see Figure 2.2). Comprehended literally, given that sCort concentration is indicative of HPA axis activity and sAA indicates ANS activity, the additive model suggests that a symmetrical association between sCort concentration and sAA activity, elevates the likelihood of challenging behaviour occurrence.

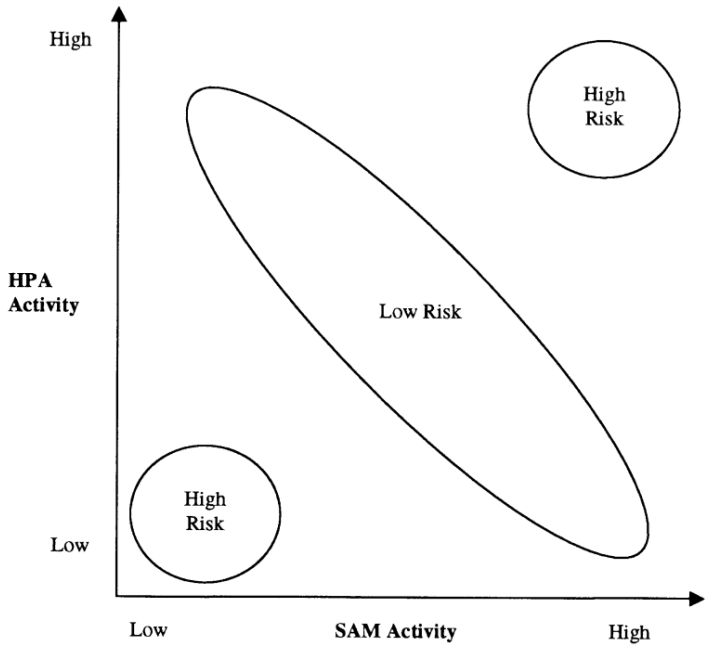


Figure 2.2 The Additive Model cited from A. M. Bauer, J. A. Quas, and W. T. Boyce, 2002, "Associations Between Physiological Reactivity and Children's Behavior: Advantages of a Multisystem Approach," *Journal of Developmental and Behavioral Pediatrics, 23*, 2, p. 106. Copyright 2015 by Wolters Kluwer Health, Inc. (see Appendix Z).

Empirical support for the validity of the additive model proposed by Bauer et al. (2002) ensued. Gordis, Granger, Susman, and Trickett (2006) assessed the validity of the additive model between two purposefully selected groups of adolescents aged between 10 and 14.4 years. The experimental group of 35 adolescents were selected on their histories of being abused and frequently exhibiting challenging behaviours. The control group was made up of 32 age-matched participants without such histories and behaviours. All participants completed two tasks: the verbal completion of a story and mathematical calculations. Saliva samples were collected before and at four times-points after the tasks. The saliva samples were analysed for sCort concentration and sAA activity as indicators of HPA axis and ANS levels. The 23-item Reactive-Proactive Aggression Questionnaire developed by Raine et al. (2006) was completed by the participants and their primary carers for data regarding challenging behaviour. The authors concluded that higher rates of aggression reported by primary carers were recorded when a symmetrical association between low sCort concentration and sAA activity was detected.

Further, El-Sheikh, Erath, Buckhalt, Granger, and Mize (2008) assessed the validity of the additive model among 64 children with an average age of 8.72 years.

The authors applied an experimental treatment design where the participants listened to an audio recording of an argument between an adult male and female. The outcome measures for the study were sCort concentration, sAA activity and child adjustment as rated by primary carers via the second edition of the Personality Inventory for Children designed by Lachar and Gruber (2001). The authors reported that a symmetrical association between sCort concentration and sAA activity resulted in a higher primary carer rating of maladjustment therein supporting the additive model.

The interactive model as published by Bauer et al. (2002) was suggested as an alternative to the additive model. This model predicted that the risk of challenging behaviour expressed by children was higher when the association between the HPA axis and ANS was asymmetrical (Bauer et al., 2002; Koss et al., 2014). Conversely, the model predicted that the risk of a child expressing challenging behaviour was lowest when the association between the HPA axis and ANS was symmetrical (see Figure 2.3). Regarding the biomarkers of arousal, the Interactive Model suggests that an asymmetrical association between sCort concentration and sAA activity elevates the likelihood of a challenging behaviour occurring.

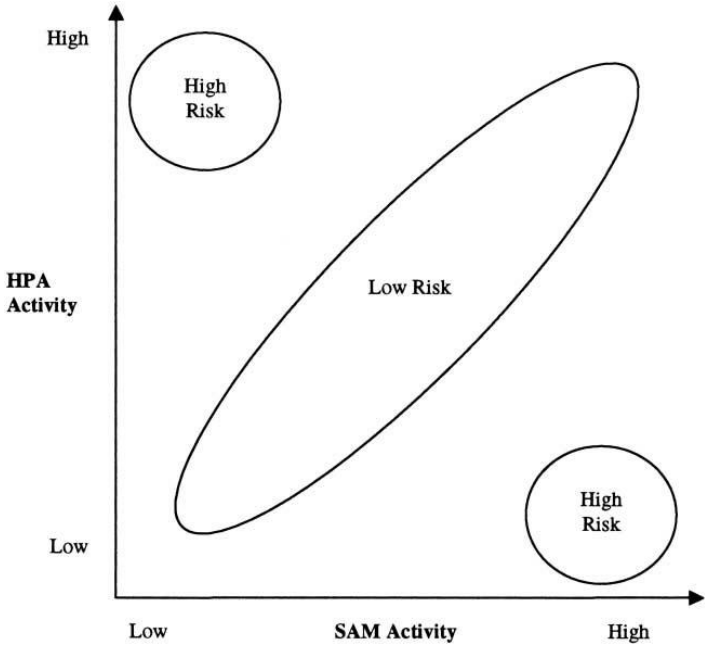


Figure 2.3 The Interactive Model cited from A. M. Bauer, J. A. Quas, and W. T. Boyce, 2002, "Associations Between Physiological Reactivity and Children's Behavior: Advantages of a Multisystem Approach," *Journal of Developmental and Behavioral Pediatrics, 23*, 2, p. 106. Copyright 2015 by Wolters Kluwer Health, Inc. (see Appendix Z).

Numerous studies have published results which corroborate the interactive model. Vigil, Geary, Granger, and Flinn (2010) investigated potential associations between sCort concentration, sAA activity and psychological well-being via the self-report Aggression Questionnaire (Buss & Perry, 1992) which included aggressive tendencies amongst two groups of children. The experimental group was composed of 62 children with an average age of 14.9 years who were residing in a family relocation facility two months after surviving the natural disaster Hurricane Katrina in 2005. Matched by age, ethnicity and socioeconomic status, the control group contained 53 children who lived in the state of Missouri in the USA. The authors reported finding that an asymmetrical association of lower sCort concentration and higher sAA activity predicted a higher risk of aggressive behaviour amongst the experimental group compared to the control group.

In addition, Allwood, Handwerger, Kivlighan, Granger, and Stroud (2011) investigated the validity of the interactive model amongst 56 children aged 7 to 16 years. The authors collected saliva samples for the analysis of sCort concentration and sAA activity. Additionally, the Revised Children's Manifest Anxiety Scale (RCMAS) designed by Reynolds and Richmond (1978/2000) and the Child Behavior Checklist 6-18 (CBCL; Achenbach, 1991; Achenbach & Rescorla, 2001) were administered. Behavioural data were collected from all participants before and after completing tasks which elicited a peer rejection scenario. The authors identified that an asymmetrical association between sCort concentration and sAA activity resulted in more challenging behaviour and internalising tendencies; therein supporting the interactive model theory.

Despite the published support for both models suggested by Bauer et al. (2002), research affirming an association between the HPA axis and ANS has been inconsistent. Some authors suggest that no association exists between the two systems (Lisonbee, Pendry, Mize, & Gwynn, 2010; Spinrad et al., 2009). In support of this contention, the way in which humans respond behaviourally and biologically to elevated arousal can depend on multiple factors such as the unfamiliarity of the experience, age, gender, environment and socioeconomic status (Corbett et al., 2012). Regarding children, the arousal response can be impacted by developmental achievements, the type of stressor, poor sleep and exposure as well associated historical factors (Chicoine et al., 2013; Koss et al., 2014). Indicatively, there remains a need to assess potential associations between the HPA axis and ANS systems and

how these systems effect behaviour (Bauer et al., 2002). Furthermore, the way in which the HPA axis and ANS interact, integrate and influence each other is not yet categorically known (Koss et al., 2014).

2.11 Summary

Children with ASD are vulnerable to exhibiting SIB and research focussing on treatment is much needed (Horner et al., 2002). Of concern, the developmental trajectory of SIB remains largely unknown (Koenig, Thayer, & Kaess, 2016). Further, there is an ever-present need to determine "what is known and not known" about challenging behaviour exhibited by young children (Conroy, Dunlap, Clarke, & Alter, 2005, p. 158). Regarding SIB, "When these behaviours are present, they should be treated" (Matson & Nebel-Schwalm, 2007, p. 573). Ultimately, SIB can be detrimental to educational development, social opportunities and threaten the lives of children with developmental disabilities such as ASD (Machalicek et al., 2007; Richman, 2008). Treating SIB presents significant challenges for practitioners, especially when the behaviour endures (Barrera et al., 2007; Horner et al., 2002; Iwata, Dorsey, et al., 1994). Without adequate research and treatments, the prognosis for children with challenging behaviours such as SIB can be bleak (Campbell, 1995).

Lourie (1949) was the first to suggest that SIB may function to regulate arousal for people with developmental disabilities. Although a universal cause for SIB is currently unknown, it is reported to have an underlying biological component (Canitano & Scandurra, 2011). This underlying component may be the reported link between SIB and increasing levels of arousal (Jennett, Hagopian, & Beaulieu, 2011). Biological data such as sCort and sAA can provide indications of internal reactions to external experiences (Kandel et al., 2000). Symons et al. (2003) reported that high sCort concentrations have been linked to SIB in adults with developmental disabilities such as ASD. Arousal has been reported to increase prior to SIB and elicit a sense of relief after SIB for adults with developmental disabilities including those with LFA (Barrera et al., 2007; Haines, Williams, Brain, & Wilson, 1995a, 1995b; Jones, Congiu, & Stevenson, 1979). Music Therapy has been reported to reduce disruptive behaviours amongst children with ASD and may be beneficial for reducing arousal (Avers et al., 2007; Boso, Emanuele, Minazzi, Abbamonte, & Politi, 2007; Hooper et al., 2012;Mra´zova´ & Celec, 2010).

Ford (1999) published a single-subject research design in the *Journal of Music Therapy* designed to test the efficacy of four treatment modalities on teeth-grinding

SIB, mouth-scratching SIB and head-hitting SIB exhibited by a 23-year-old female with severe and comorbid developmental disabilities. An initial cohort of three participants were observed for 2 hours each day over a 2-week period for inclusion screening. As a result, a 23-year-old female with profound communication, movement, weight-bearing and cortical blindness disabilities was accepted into the treatment. The cognitive deficits of the participant as described by Ford (1999) were consistent with an ID.

The participant's teeth-grinding SIB was defined as grinding her lower teeth against her upper teeth. This SIB was reported to have resulted in the exposure of her dental nerves and the onset of a painful condition called temporomandibular muscle disorder. The participants mouth-scratching SIB was described as her using her thumb nail to scratch the roof of her mouth repetitively. This was reported to have caused eating difficulties due to unhealed internal mouth sores and scratches and substantial blood loss. The participants head-hitting SIB was defined as her using her fists to strike the rear and left portions of her head with frequent repetition and excessive force. This SIB resulted in lacerations as well as discolouration at impact sites. Prior to participating in the research, interventions designed to treat her SIB were reported to have focussed on sound, touch, visual, smell and motor sensory strategies.

Ford (1999) applied a single-subject ABACADA research design where the A condition was SIB blocking, the B condition was music listening, the C condition was water play and the D condition was active MT. More specifically, the A condition involved the researcher using her hand to physically block the impact of the SIB. For example, the researcher blocked the head-hitting SIB by placing her hand between the head and fist of the participant to ensure that no physical injury resulted from this behaviour. Condition B required the participant to listen to music administered via headphones. During condition C, the participant was assisted to engage in tabletop play with sponges and balls in a tub filled with water which was heated to body temperature. In condition D, the participant was supported to engage in free-play with an electronic piano keyboard. Conditions B, C and D were administered for seven consecutive days each. The initial A condition lasted for nine consecutive days, the second and third return A condition treatments lasted for five consecutive days each and the final Phase A continued over four consecutive days.

For each of the conditions, the participant was removed from a group activity and observed for 40 minutes, engaged in treatment for 30 minutes then observed for

an additional 40 minutes in isolation. Of these 40 minute periods, the target SIB was rated in 10 second intervals during randomly selected five minute periods. SIB percentages were calculated by dividing the observed SIB occurrences by the total number of 10 second increments filmed before being multiplied by one hundred.

Footage of behaviour during treatment phases was not recorded. However, SIB frequencies for head-hitting and mouth-scratching whilst engaged in condition C and D may have proven invalid or acted as a differential reinforcement of alternative behaviour (DRA) due to the participant requiring use of her hands to participate in the activities and engage in SIB simultaneously. To participate in the research, the participant travelled by bus from her residential accommodation for people with high support needs to the community training venue which had been built to provide services for adults with severe developmental disabilities.

Ford (1999) reported that the teeth-grinding SIB decreased from 52% (SD = 19.48) before music listening to 35.7% (SD = 24.71) after. This suggested that listening to music was effective in the reduction of this type of SIB. As such, this result concurs with other reports that music listening can be an effective method of reducing SIB for people with profound disabilities such as LFA (Matson & LoVullo, 2008). However, music listening was not reported to reduce head-hitting or mouth scratching SIB. Limitations based on inadequate environmental controls, not establishing a behavioural baseline prior and not determining the function of SIB prior via a functional analysis were reported as potential confounds by the author.

Recent literature suggests that children with ASD often use music for therapeutic purposes (Finnigan & Starr, 2010). In addition, increasing SIB has been linked to elevated arousal of a child with LFA (Jennett, Hagopian, & Beaulieu, 2011). Therefore, SIB may act as a mechanism to regulate arousal levels for individuals with intellectual and developmental disabilities such as ASD (Lourie, 1949; Novak, 2003). Furthermore, Kidd et al. (2012) reported that sCort concentration and sAA activity was higher amongst children with ASD who recorded lower intelligence quotients (IQ) scores; consistent with a diagnosis of LFA. Hence, the analysis of biological data from children with LFA may contribute to the development of more effective treatments for SIB.

CHAPTER 3: RATIONALE

3.1 Why Treat SIB for Children with LFA?

The increasing global prevalence of ASD presents the need for further research, services and effective treatments (Saemundsen, Magnússon, Georgsdóttir, Egilsson, & Rafnsson, 2013). Specific to the current research, treatments designed to reduce the physical and psychological impacts of SIB amongst school-aged children with ASD and LFA are much needed (Healey et al., 2001; Horner et al., 2002). Such treatments may also act to lessen the psychological trauma experienced by school teachers and primary carers who are tasked with managing SIB in situ (Hastings & Brown, 2002; Matson & Nebel-Schwalm, 2007). At present, two main therapeutic approaches have been reported to be effective in reducing SIB among children with LFA; pharmacological and behavioural therapies.

Pharmacological therapies designed for individuals with ASD can act to target specific maladaptive behaviours such as SIB (King & Bostic, 2006). Despite the U.S. Food and Drug Administration (FDA) Administration endorsing the use of risperidone and aripiprazole to treat SIB exhibited by children with LFA, the efficacy of these medications can be limited by adverse effects, medication resistance, existing comorbid conditions such as ID and the vast spectrum of possible ASD presentations (Adler et al., 2015; Mohiuddin & Ghaziuddin, 2013). Further, the heterogeneity of the ASD and ID components of the LFA diagnosis make medication resistance more likely (Adler et al., 2015). In instances where a person with LFA is not medication resistant, pharmacological therapies can act to treat symptoms other than the intended SIB (Williamson & Martin, 2012). The validity of peer-reviewed literature supporting pharmacological therapies designed to treat SIB amongst children with ASD is often limited by small sample sizes, low replicability, and short trial durations (Benvenuto et al., 2013).

Matson, Sipes, et al. (2011) published numerous treatment recommendations as a potential framework for reduction of challenging behaviours such as SIB for adults with ASD. The authors recommended that an assessment of SIB function be incorporated in targeted research to form an evidence-based approach. The Motivation Assessment Scale (MAS) published by Durand and Crimmins (1988) has been reported to be a valid instrument for such as purpose. The MAS enables primary carers to report the function of challenging behaviours by eliciting responses to determine

whether the behaviour results in attention, tangible objects, avoidance or sensory inputs (Joosten & Bundy, 2010). To augment the MAS, and as previously reported, determining the function of SIB via a FA is an essential component of the treatment process. Matson, Sipes, et al. (2011) also recommended that challenging behaviour such as SIB be assessed before and during interventions to enable a comprehensive and complete analysis. Finally, the authors recommended that a collaborative and multidisciplinary approach be adopted where possible for the formulation of individualised and targeted treatments for challenging behaviours such as SIB.

Applying recommendations such as the above-mentioned to behavioural treatments designed to reduce SIB for children with LFA may help improve functional independence for participants in both therapeutic and educational settings (Smith et al., 2005). Further, involving natural supports such as primary carers in such treatments may be advantageous, although for this to occur, empirically established methods of treatment are required (Richman, 2008).

3.2 Music Listening

A form of behavioural therapy, music listening may lead to promising results in the treatment of young children with developmental disabilities such as LFA (Conroy et al., 2005). Music is easy to apply, non-invasive, comes at minimal financial expense, and can help in the management of anxiety amongst children, including those with ASD (Avers et al., 2007). Specific to the present research, children with LFA have been reported to have the capacity to participate in music listening regardless of motor or cognitive abilities (Ford, 1999).

Whipple (2004) reported that music therapy studies designed to treat children and adolescents with ASD often contain methodological issues and lack quantitative data to support their findings. Therefore, more empirically valid evidence is needed to further establish the validity and generalisability of music therapy (Hillecke, Nickel, & Bolay, 2005). Functionally, children with severe disabilities such as LFA have been known to refuse wearing MP3 music players or tolerate wearing ear phones (Hooper et al., 2012). Despite this, it is known that listening to music at high volume with fast tempi and uneven melodies can be excitatory. Conversely, listening to music at low volume with slow tempi and an even melody can result in calmness and a reduction in arousal (Scherer & Zentner, 2001 as cited in Fancourt et al., 2014). For example, the arousal reducing qualities of listening to classical music have been well documented (Dibben & Williamson, 2007; Holland, 1995; Khalfa et al., 2003; Suda, Morimoto,

Obata, Koizumi, & Maki, 2008). Moreover, music-based interventions which employ rigorous scientific methods for the treatment of people with ASD have the potential to be both informative and therapeutic (Boso, Politi, Barale, & Emanuele, 2006; Gold, Wigram, & Elefant, 2006; Lopez, 2009).

As an inexpensive and non-intrusive therapeutic method, music listening can be easily applied and take up minimal cognitive energy for the listener (Hooper et al., 2010; Mra´zova´ & Celec, 2010). Well-designed music-based interventions have the potential to make significant contributions to the disciplines of science and clinical practice (Lange & Lainhart, 2009). Further, the publication of effective and rigorous music-based interventions may help music therapies become a "gold standard" in clinical practice (Brandes, 2009, p. 100).

3.3 Music Listening and Arousal

Music listening has been reported to reduce sCort concentrations in adults after being exposed to a TSST (Khalfa et al., 2003). However, the physiological effects of music listening have gone underreported despite it being shown to reduce biomarkers of arousal (Cervellin & Lippi, 2011; Khalfa et al., 2003; Suda et al., 2008). In addition, listening to calming music has been reported to reduce arousal (Fukui & Yamashita, 2003; Khalfa et al., 2003; Knight & Rickard, 2001; Nilsson, 2009; Nilsson, Unosson, & Rawal, 2005).

Research designed to assess arousal has the potential to benefit both the body of knowledge and clinical practice for individuals and groups, especially for those with ASD (Kudielka & Wüst, 2010). More specifically, assessing biomarkers of arousal such as sCort and sAA amongst those with ASD can be highly relevant as the baseline arousal levels of this cohort are often markedly elevated (Bitsika et al., 2014; Jessop & Turner-Cobb, 2008). Research with this focus can produce valuable information regarding treatment efficacy, in addition to within and between-group variance associated with ASD (Anderson, 2015).

The analysis of arousal biomarkers such as sCort concentration and sAA activity can present an unambiguous measurement of the human body's internal state (White & Mulligan, 2009). Specific to behavioural therapies such as music listening, these biomarkers form a rigorous and multiple systemic approach to the measurement and analysis of arousal (Bauer et al., 2002; Filaire, 2009; Fortunato et al., 2008; Monteleone et al., 2011; Nigg, 2006).

3.4 The Hypothalamic-Pituitary-Adrenocortical Axis and Cortisol

The analysis of sCort concentrations can provide a hormonal indicator of the HPA axis and yield a reliable measurement of the human arousal (Hellhammer, Wüst, & Kudielka, 2009; Viau, 2010). However, autoimmune disorders such as asthma and atopic dermatitis, medications prescribed for sexual reproduction, glucocorticoid receptor gene polymorphisms and unpredictable external psychological stressors can affect cortisol readings (Kudielka, Hellhammer, & Wüst, 2009; Lanni et al., 2012). Further, sCort data collected from post-pubescent females must be interpreted with caution due to potential variations resulting from the menstrual cycle (Bitsika et al., 2014). Given that cortisol levels can be influenced by numerous individual and naturally occurring factors, standardising saliva collection time-points is seen as best practice for accurate results (Bauer et al., 2002; Chicoine et al., 2013). Of use to research, a standard morning diurnal peak in sCort concentration can provide an ideal opportunity to assess treatment effect when the activity is at its highest point (Fries et al., 2009; Hellhammer et al., 2009; Kudielka & Wüst, 2010). Further, changes in sCort concentrations are typically most evident between 10 and 20 minutes after exposure to an arousing event or stimuli (Corbett et al., 2012; Quas, 2011).

Accounting for the above-mentioned factors, the analysis of cortisol can yield objective data which can be indicative of arousal regulation (White & Mulligan, 2009). In addition, the cortisol activity of people with ASD have been reported to be comparable to those who develop typically (Corbett et al., 2006; Lam, Aman, & Arnold, 2006). Despite this, there is an ongoing and specific need to investigate the HPA axis and cortisol activity in children in order to develop methods to reduce arousal (Jessop & Turner-Cobb, 2008).

Specific to the present research, higher sCort concentration has been linked to SIB exhibited by adults with developmental disabilities (Symons et al., 2003). However, this result has not yet been replicated amongst school-aged children with LFA. In addition, Bitsika et al. (2015) suggested that future research be conducted into the legitimacy of cortisol as an arousal biomarker for children with ASD to contribute the body of knowledge in this field.

3.5 The Autonomic Nervous System and Alpha-Amylase

Biological and behavioural research designs can include an analysis of sAA because it offers an accurate, reliable and non-invasive indication of ANS activity; often most useful in single-case studies (Cheshire, 2012; Nater et al., 2005; Rohleder

& Nater, 2009; Rohleder, Nater, Wolf, Ehlert, & Kirschbaum, 2004). However, further research is needed to confirm it as an endorsed biological marker of ANS activity specifically relevant to children with ASD (Bosch, Veerman, de Geus, & Proctor, 2011; Quas, 2011). For example, children with ASD can change biologically from being under-aroused to over-aroused as they attempt to regulate states of relaxation or alertness, the causes of which remain largely unknown (Joosten, Bundy, & Einfeld, 2009; Ozsivadjian, Knott, & Magiati, 2012). In addition, measures of sAA activity are dependent on numerous extraneous factors such as saliva gland function, parasympathetic and sympathetic influences on protein secretions and saliva flow rate (Cheshire, 2012).

However, indicators of ANS activity such as sAA have the potential to provide a measure of the internal and external drivers of challenging behaviour such as SIB (Nigg, 2006). Therefore, there is a current need to conduct further ANS investigations including sAA analysis amongst children with ASD (Kidd et al., 2012). Single-case designs may provide an appropriate starting point to assess sCort and sAA on an individual basis prior to group replication.

3.6 Group or Single-Case Research

Group research designs, such as randomised controlled trials (RCTs), are commonly used to study the effects of interventions for children with ASD (Kasari & Smith, 2013). With appropriate numbers of participants for statistically powerful analyses, these designs can enable the systematic assessment of treatment effects and the potential identification of underlying behavioural mechanisms and mediators (Kasari & Smith, 2013). Further, RCTs allow researchers to account for participant variables within groups which can confound results by type 1 error (Smith et al., 2007). Even though group research designs featuring greater numbers of participants enable the assessment of treatment efficacy over longer periods, single-case research designs enable the assessment of treatments under more varied and naturalistic conditions and present opportunities to detect behavioural mediators and moderators (Smith et al., 2007).

Single-case research involves the participant acting as their own control in treatments designed to create behaviour change (Kasari & Smith, 2013). Specific to the present research, such designs can be used in the treatment of school-aged children with ASD (Machalicek et al., 2007). Although the results of single-case research can lack immediate population generalisability, when a functional association is detected

through the application of a rigorous methodology, greater confidence can be attributed to findings (Finnigan & Starr, 2010). Single-case designs are often used when participants require tailored treatments and treatment efficacy is deemed beneficial prior to including larger samples (Shadish, 2014). Further, Fisherian statistical analyses such as the Fisher's Exact tests are suggested appropriate for use when analysing data from single-case designs (Perone, 1999).

Chambless and Ollendick (2001) reported that empirically supported treatments designed for children can be effective in clinical environments. More specifically, interventions that scientifically assess challenging behaviours exhibited by children with ASD via behavioural observation can have a high clinical relevance (Matson & Nebel-Schwalm, 2007). For example, Woodyatt, Marinac, Darnell, Sigafoos, and Halle (2004) recorded footage of children with severe IDs in familiar environments so that observers, who had been trained by the authors, could rate the incidence of SIB in 10-second intervals. Similarly, Singh and Winton (1984) also reported assessing the incidence or non-incidence of SIB at 10-second intervals. The 10-second interval rating method, known as partial interval recording (PIR), can be used in the assessment of SIB intervals exhibited by children with ASD (Matson & Nebel-Schwalm, 2007). Regarding these behavioural observations, Matson, Sipes, et al. (2011) suggested that they be conducted by at least two independent raters who possess appropriate qualifications and training to ensure the establishment of inter-rater reliability. Moreover, visual observation methods such as PIR producing frequencies is the most commonly used and can be an appropriate method of analysing behaviour within single-subject research designs (Horner et al., 2005).

3.7 The Three-Stage Mediating Model

From a physiological perspective, children's arousal regulation depends on the individual child and the environment within which they interact (Bauer et al., 2002). Specifically, those with ASD who do not respond typically to demand conditions can be especially vulnerable to stress regulation difficulties and exhibit the most challenging of behaviours (Kidd et al., 2012). Valuable information about children's abilities to regulate arousal can arise from single-case research designed to assess physiological stress reactions (Bauer et al., 2002). Indicatively, "proper regulation is a key feature of the stress-response pathways" (Kidd et al., 2012, p. 2657). A specific or universal link between SIB and ASD is yet to be established (Benvenuto et al., 2013).

Hence, the aim of the present research was to apply three studies designed to assess the validity of a theoretical three-stage mediating model (see Figure 3.1). Specifically, the mediating model assesses the potential for biological arousal via sCort concentrations and sAA activity to mediate a relationship between music listening and SIB for school-aged boys with LFA after being exposed to both simulated and actual morning school bus rides. The three components of the mediating model (music listening, arousal, and self-injurious behaviour) align in a stepwise fashion with the three studies described and ultimately culminate in determining the validity of the theoretical model.

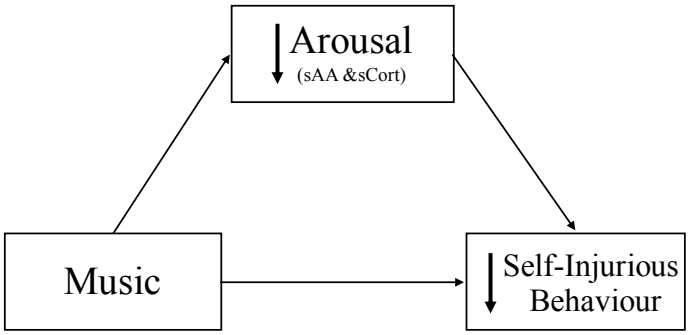


Figure 3.1 The three-stage mediating model.

3.7.1 Study 1

Selecting music for music-based therapies designed to treat people with LFA is often made more difficult by communicative and cognitive difficulties associated with the condition (Hooper et al., 2010). However, in the later part of the 1960s through to the early 1970s, a seminal author in the treatment of SIB, Ivar Lovaas was one of the first to welcome primary carers into treatments for their children to assist with communication (Smith & Eikeseth, 2011). More recently, Maglione, Gans, Das, Timbie, and Kasari (2012) suggested that interventions for children with ASD should include primary carers.

Similarly, it is not uncommon for primary carers of children with ASD to rate their child's anxiety (Bitsika et al., 2014). Therefore, when people with an ID such as those with LFA experience difficulties in selecting their own calming music for anxiety reduction, those without an ID such as their primary carers can meaningfully select on their behalf (Hooper et al., 2010).

Study 1 represents the music listening component of the theoretical model. This study aimed to address a gap in the literature regarding the selection of music for

music-based interventions by incorporating recently published factors about calming music, musical composed within specific forms (Rondo, Theme and Variations, Sonata), and an initial music playlist selection, selected by a world renowned concert pianist and parents/primary carers for their children with LFA as music raters. The establishment of this method of selecting music for music-based interventions resulted in the selection of a single calming music segment, a music selection methodology for future research and music for use in the following two studies. It was hypothesised that employing a music selection method and parents/primary carers' ratings would identify a single most calming musical segment for music listening in Study 2 and Study 3.

3.7.2 Study 2

Study 2 assessed the possible link between the music listening and arousal components of the theoretical model and tested the potential for music listening to reduce salivary biomarkers of arousal among school-aged males with LFA both before and at four time points after a simulated morning school bus ride. The school bus simulator, an adaptation of the TSST-C, was designed and constructed to imitate a naturalistic morning school bus ride for the participants who were regular school bus users. The realism of the school bus simulator bus was boosted by the use of visual and audio recordings taken from real morning school bus rides as well as the inclusion of a genuine school bus seat pair, a surround sound stereo system with exact screen dimensions and sitting distance as was apparent in the real school bus. Not only did this design create an accurate imitation of a morning school bus ride, but it prevented simulator sickness which may have been apparent for participants' whilst in the simulator. Experimental and control group saliva samples were collected from participants before and after the school bus ride simulation. The final stage of the research was to assess this in a naturalistic environment and the potential for it to reduce SIB. It was hypothesized that boys with LFA who listened to calming music whilst being exposed to a school bus simulator as a modified TSST-C would record significantly lower sCort concentrations and sAA activity than those randomly allocated to the no music group.

3.7.3 Study 3

A culmination of the music listening, arousal and SIB components of the theoretical three-stage mediating model, Study 3 assessed the potential for biological arousal to mediate a relationship between music listening and SIB attempt frequencies

amongst three school-aged males with LFA. The term SIB attempt frequencies indicates that participants were required to wear personal protective equipment as prescribed for SIB harm-minimisation prior to the commencement of the research. A single-case design comprising two conditions with repeated trials, Study 3 assessed the validity of the three-stage mediating model on an actual morning school bus ride to assess its validity in a naturalistic environment. In addition, the motivating factors associated with each participant's SIB were assessed via the Motivation Assessment Scale (MAS) and combined with SIB and sCort concentration with sAA activity data to complete each single-case presentation. The four treatment days were conducted on the same day over four consecutive weeks. It was hypothesised that music listening would elicit significant sCort concentration, sAA activity and SIB frequency reductions amongst three boys with LFA on the school bus and at school.

CHAPTER 4: STUDY 1

In the absence of a standardised music selection methodology, the primary aim of Study 1 was to establish a method of selecting music for music-based interventions. The music selection method incorporated musical form, segment, purpose, online distribution, de-identification, randomisation and expert musical consultations. The aim of employing this method was to purposefully select a single piece of calming music for use in Study 2 and Study 3 as well as define the music listening component of the theoretical three-stage mediating model (see Figure 4.1). Further, the method of music selection may be considered for use in future music-based interventions.

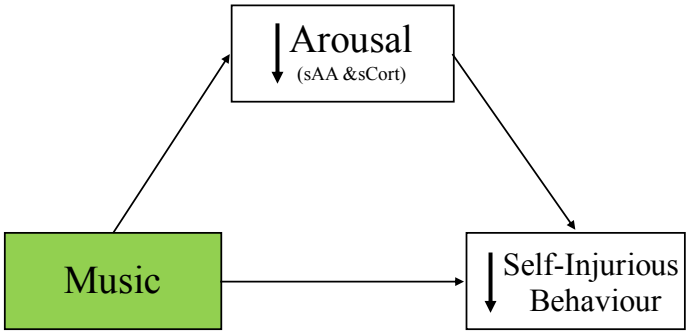


Figure 4.1 Study 1: Three-stage mediating model.

4.1 The Problem: An Absence of Music Selection Methods

Without a single standardised definition, music can be generally defined as "creative play with sound" (Brandt, Gebrian, & Slevc, 2012, p. 3). Therein, selecting music for research can be a complex and difficult task. However, failing to report methods used to select music for music-based interventions can greatly reduce the validity and generalisability of findings (Robb & Carpenter, 2009). A prime example of this was the music-based intervention published by Rauscher, Shaw, and Ky (1993): *Music and spatial task performance*. This single-page publication reported how 36 university students were exposed to the first 10 minutes of three auditory conditions prior to completing a selection of Stanford-Binet Intelligence Scale spatial-reasoning tasks. The first condition involved listening to Wolfgang Amadeus Mozart's sonata for two pianos in D major (K488), the second listening to a relaxation recording and the third silence. Raw spatial-reasoning test scores for each participant were standardised and used to calculate an overall IQ. Based on these scores, the authors reported that listening to K488 resulted in significant improvements in spatial reasoning. However, a justification for selecting K488 study was not reported; presenting a methodological

limitation to the validity of the dependent variable. Despite this oversight, and based on the results of the afore-mentioned study by Rauscher et al. (1993), a promoter from the USA named of Don Campbell began to market numerous learning and health benefits of listening to music composed by Mozart; known in the media as *The Mozart Effect* (Campbell, 1997).

This marketing campaign attracted considerable public, corporate, media and scientific attention (Crncec, Wilson, & Prior, 2006; Nantais & Schellenberg, 1999). Books, CD's and videos were used to promote the idea that listening to Mozart's music could elicit relaxation, healing and boost the intelligence and learning capacities for children and adults (Campbell, 1997; Crncec et al., 2006; McKlevie & Low, 2002). So successful was Campbell's promotion, Governor Zell Miller for the state of Georgia in the US budgeted "a compact disc or cassette for each infant born in the state" to listen to Mozart's music (Nantais & Schellenberg, 1999, p. 370).

Despite the alleged benefits marketed by Don Campbell, the original research findings by Rauscher et al. (1993) were not then, and have not since, been sufficiently replicated (Crncec et al., 2006; Nantais & Schellenberg, 1999). As such, music-based interventions have become thought of as treatments of last resort in the scientific community (Brandes, 2009). Unfortunately, the absence of a methodological justification for the selection of music in the Rauscher et al. (1993) study is apparent within the wider music-based intervention literature. Justifications for music selection are absent in meta-analyses, systematic reviews, critical examinations and a Cochrane review (Accordino, Comer, & Heller, 2007; Brown & Jellison, 2012; Gold, Voracek, & Wigram, 2004; Gold et al., 2006; Hooper, Wigram, Carson, & Lindsay, 2008; Simpson & Keen, 2011; Whipple, 2004). In addition, music-based interventions often neglect to report music titles and/or the form in which music was composed (Lanovaz, Fletcher, & Rapp, 2009; Rapp, 2007; Stephens, 2008), the qualifications of those whom selected the music (Carnahan, Basham, & Musti-Rao, 2009; Corbett, Shickman, & Ferrer, 2008; Simpson & Keen, 2010), include music selected by an uncredentialed panel, or limit participant music choice to playlists chosen without qualification by the authors (Crncec et al., 2006; El Hassan, McKeown, & Muller, 2009; Lesiuk, 2008). Of concern, the validity of peer-reviewed music-based interventions results appears to be compromised. This appears to be the case in music-based interventions designed to treat children with ASD.

Katagiri (2009) included a panel of four highly credentialed musicians and inter-rater reliability to match musical melody, lyric, tempo, volume and articulation to the emotions of children with ASD. However, the use of "Japanese children's songs designed to teach emotions" without further detail appeared to lack adequate specificity (Katagiri, 2009, p. 21). Pasiali (2004, pp. 14,15) asked primary carers of children aged seven to nine years with ASD to help create songs to match the melodies of *You Are My Sunshine* and *Yellow Submarine*. Kern, Wakeford, and Aldridge (2007, pp. 45,46) reported the specific titles of music used: *The Wheels On The Bus Go Round And Round*; *Swing Low, Sweet Chariot*; *Ring Around The Rosy*; and *Row, Row, Row Your Boat* in a music-based intervention for a three-year-old boy with ASD. Dellatan (2003, p. 107) reported the use of music from a specific CD entitled *We Sing Children's Songs* to treat a five-year-old boy with ASD. Orr, Myles, and Carlson (1998, p. 164) reported the use of music from a CD entitled *Rhythmic Medicine* in a music-based intervention for an 11-year-old girl with ASD.

Identifying a method of music selection for ASD music-based interventions would consolidate the above-mentioned attempts while improving the internal and external validity of results. To build scientific rigor around such a method, it may be useful to consider the musical period, composers and forms of potential music for selection.

4.2 Selecting Music by Period and Composer for Music-Based Interventions

The Baroque (1600 to 1750), Classical (1750 to 1830), Romantic (1830 to 1860), and Post-romantic (1860 to 1920) periods of musical composition produced some of the most famous musical works created by some of the most well-known composers (Dalla & Peretz, 2005, p. 66). Selecting music by period for music-based interventions can add methodological rigour by narrowing the musical scope to a certain period for more specific identification of the independent variable. In doing so, the designs would be more easily replicated and results more easily testable.

The use of classical music in music-based interventions has been well documented (Dibben & Williamson, 2007; Gerra et al., 1998; Khalfa et al., 2003; Suda, Morimoto, Obata, Koizumi, & Maki, 2008). Further, selecting music by period, selecting music by composer can further refine the independent variable. For example, within the Classical period, Wolfgang Amadeus Mozart, Ludwig van Beethoven and Johann Sebastian Bach were reported to have produced the greatest number of and most well-recognised compositions (Murray, 2003 as cited in Kozbelt, 2009). Alive

from 1756 and 1791, Mozart composed approximately 10,136 pieces over a period of around 31 years. Beethoven lived from 1770 to 1827 during which time he composed an estimated 4,902 pieces in a career spanning approximately 45 years. Music composed by these artists within the classical period can be easily identified by the structure within which it was composed: otherwise known as its musical form. This method would further specify the method of music selection and enable easier replication for result validation. In addition to specifying the selection of music by period and composer, music selection by form may act to further clarify the independent variable within music-based interventions and improve replicability.

4.3 Common Musical Forms of the Classical Period

The continuity, balance and shape within which music is composed is known as musical form (Bennett, 1980). Music composed in rondo, theme and variations and sonata forms by Mozart, Beethoven and Bach can be readily distinguished by their structure and appeared frequently in compositions of the Classical period.

The rondo musical form is identified by a repeating theme within intermittent contrasting episodes (see Figure 4.2; Abdy-Williams, 1890; Bennett, 1980; Randel, 2003). Examples of music composed in Rondo form are Wolfgang Amadeus Mozart's rondo for piano No. 3 in A minor, K. 511 and adagio for glass harmonica in C major, K. 356 (K. 617a).

Phase	A	B	A	C	A	D	A
Theme	Principal	2nd	Principal	3rd	Principal	4th	Principal

Figure 4.2 Study 1: Rondo musical form.

The theme and variations form progressively varies the harmony, tempo or rhythm of an original theme (see Figure 4.3; Bennett, 1980; Randel, 2003). Examples of music composed within this musical form include Wolfgang Amadeus Mozart's variations for piano (12) in C major on 'Ah, vous dirai-je maman' K. 265 (K. 300e) and Johann Sebastian Bach's Goldberg Variations, BWV 988- variation 13(2).

Phase	A	A'	A''	A'''	A''''	CODA
Theme	Theme	1st Variation	2nd Variation	3rd Variation	4th Variation	Conclusion

Figure 4.3 Study 1: Theme and variations musical form.

Music composed in sonata form establishes a main theme known as the Exposition, modifies the theme to create tension known as the Development, resolves the tension in the Recapitulation and concludes the structure in what is known as the Coda (see Figure 4.4; Ordway, 2008; Randel, 2003). Some examples of music composed in sonata musical form are Ludwig van Beethoven's Sonata No. 8 in C minor, Op. 13 Adagio cantabile and Sonata No. 14 en do dièse mineur, Op.27, No 2 adagio sostenuto.

Phase	EXPOSITION	DEVELOPMENT	RECAPITULATION	CODA
Theme	Principal/Bridge/Second/Closing		Principal/Bridge/Second/Closing	

Figure 4.4 Study 1: Sonata musical form.

Considering the above-mentioned musical periods, composers and forms provides clarity to the independent variable applied to music-based interventions. This is likely to result in easier replication and greater internal and external validity. As such, a standardised music selection method for music-based interventions does not currently exist (Robb & Carpenter, 2009). In addition, music-based interventions should at least meet minimum contemporary scientific methodological standards (Bradt, 2012). The following method of music selection endeavours to meet these standards.

4.4 Method

4.4.1 Participants

4.4.1.1 Study 1: A

With verbal information provided by the researcher as to the purpose of Study 1, two primary school music teachers were consulted to determine the most appropriate forms within which to choose music for the study. Both music teachers had in excess of ten years' experience teaching music to school-aged boys with disabilities including LFA.

A concert pianist was engaged to select the music for a calming music playlist in compliance with a specific music selection framework discussed in the procedure section. The pianist had achieved numerous national and international accolades and produced several gold rated albums.

4.4.1.2 Study 1: B

Primary carers who rated the calming properties of the music contained within the calming music playlist were recruited via electronic mail sent by the researcher to government and non-government agencies who provided support services to those with LFA (see Appendix M). Of the 192 primary carers who opened the survey, 77 self-identified as a primary carer for a male with LFA and 60 provided details of their country and state of origin. Of the 60 who provided details of their country of origin, 43 reported being from Australia, seven from the USA, four from the UK, four from Canada, and one each from Rome, France, Poland and Estonia.

Eighteen primary carers self-reported possessing formal music training in various instruments such as the clarinet, guitar, violin, trumpet, piano and double bass of between 1 and 10 years. This music training was reported to range from high school level to board certified and registered music therapy level inclusive of Bachelor and post graduate qualifications. Of the 60 primary carers who recorded their country and state of origin, a total of 32 of the 48 primary carers who consented to participating completed the survey.

4.4.2 Procedure

The Curtin University Human Research Ethics Committee granted ethical approval for this and Study 2 and Study 3. This was based on compliance with the *National Statement on Ethical Conduct in Humans Research* (NHMRC) (National Health and Medical Research Council, 2007) and that the researcher was in the possession of a current *National Police Clearance* and *Working with Children Check Card*. Study 1, Study 2 and Study 3 were assigned the ethics approval number of HR138/2011, which appeared on all correspondence along with the contact details of the researcher, primary supervisor and ethics committee (see Appendix M, N, O, P, Q, R).

4.4.2.1 Study 1 - A

The researcher consulted with one male and one female primary school music teacher to determine the initial elements of a calming music playlist framework which was to be sent to the concert pianist for music selection. The age of the teachers was 46 and 41 years respectively. They both reported having in excess of 10 years' experience teaching music to primary school children with disabilities. The teachers confirmed that music composed within the classical period was most appropriate as had previously been established (Dibben & Williamson, 2007; Gerra et al., 1998;

Khalfa et al., 2003; Suda, Morimoto, Obata, Koizumi, & Maki, 2008). Further, the teachers confirmed that selecting music composed within the well-known forms of Rondo, Theme and Variation and Sonata for the calming music playlist would result in dependent variable clarification and ease of music identification for the current study and future research. As a result, music composed within the Classical Period and Rondo, Theme and Variations and Sonata musical forms were included.

To add elements to the music selection framework, the researcher conducted a literature review in the search for descriptions of calming music. As such, "description of sedative music" which had been based on the "principle elements of musical composition" published by Sadie, 2001 as cited in Hooper et al. (2010, p. 25) was sourced and included (see Table 4.1).

Table 4.1

Study 1: Description of Sedative Music

Musical Factor		Description
Tempo		Remaining stable with gradual increases (accelerandos) or decreases (ritardandos)
Volume		Remaining stable with gradual increases (crescendos) or decreases (diminuendos)
Texture		Remaining stable with subtle changes in style or instrumentation
Melody		
	Line 1	Repetition of material.
	Line 2	Little embellishment, no unexpected pauses or breaks.
	Timbre	Gentle sound with gradual changes within and between musical families.
	Pitch	Gradual changes between registers
	Accents	Few – used to add expression rather than energy to a melodic line
Harmony		Modulations, cadences that do not introduce unexpected harmonies or dissonance

Note. Table cited from "The Practical Implication of Comparing How Adults With and Without Intellectual Disability Respond to Music," by J. Hooper, T. Wigram, D. Carson, and B. Lindsay, 2010, *British Journal of Learning Disabilities, 39,* 1, p. 25. Copyright 2015 by John Wiley & Sons Inc. (see Appendix Z).

The researcher included two additional elements to the framework. Firstly, the concert pianist would be asked to select music based on the first two minute segments only. This would ensure that all playlist music would be of equal length, therein, equally likely to be chosen as most calming. Further, reducing the length of the music

would encourage the music raters to complete the survey as it would take less time in total. Additionally, the music was to be generated by a single instrument to avoid "hyper- or hyporeactivity to sensory input" as may have been associated with the ASD component of a LFA diagnosis (see Appendix E; American Psychiatric Association, 2013, p. 51).

Once established, an outline of the calming music playlist framework was sent via email to the concert pianist. The pianist was asked to apply the framework in addition to his musical knowledge and experience to select the six 2-minute musical segments. A total of six segments were included to enable the testing of a generalised linear mixed model (GLMM) on the ratings. The concert pianist then returned the researcher's email with the titles of the six compositions (see Table 4.2). Upon receipt of the musical titles, the researcher assessed the validity of the playlist by listening to the segments to ensure they met the sedative framework published by Hooper et al. (2010). Further, the actual music titles were reviewed to ensure that they had been composed in the requested musical forms.

Table 4.2

Study 1: Calming Music Playlist

Musical form	Composer, Title and Duration
Rondo	Wolfgang Amadeus Mozart - Rondo for piano No. 3 in A minor, K. 511 (10:53)
	Wolfgang Amadeus Mozart - Adagio for glass harmonica in C major, K. 356 (K. 617a) (3:26)
Theme and Variations	Wolfgang Amadeus Mozart - Variations for piano (12) in C major on 'Ah, vous dirai-je maman' K. 265 (K. 300e) (14:00)
	Johann Sebastian Bach's Goldberg Variations, BWV 988-Variation 13(2) (2:38)
Sonata	Ludwig van Beethoven's Sonata No. 8 in C minor, Op. 13 - 2. Adagio cantabile 1 (6:24)
	Ludwig van Beethoven's Sonata no. 14 en do dièse mineur, op.27, no2 - 1 Adagio sostenuto (7:43)

Upon meeting the music selection framework, the researcher sought high quality recordings of the pieces. Ludwig van Beethoven's Sonata no. 14 en do dièse mineur, op.27, no2 - 1 Adagio sostenuto known as his *Moonlight Sonata* and Ludwig van Beethoven's Sonata No. 8 in C minor, Op. 13 - 2. Adagio cantabile 1 known as the *Sonata Pathetique* were sourced from *Your 100 favourite piano masterpieces as voted*

by listeners of ABC Classic FM (Australian Broadcasting Corporation, 2005). Wolfgang Amadeus Mozart - Variations for piano (12) in C major on 'Ah, vous dirai-je maman' K. 265 (K. 300e), Wolfgang Amadeus Mozart - Rondo for piano No. 3 in A minor, K. 511 and Wolfgang Amadeus Mozart - Adagio for glass harmonica in C major, K. 356 (K. 617a) were sourced from *Mozart: Variations; Rondo in A Minor; Adagio in B Minor* (Schiff, 1990). Johann Sebastian Bach's Goldberg Variations, BWV 988-Variation 13(2) was sourced from *A State of Wonder: The Complete Goldberg Variations, 1955 & 1981* (Gould, 2002).

The music recordings were vetted for sound quality before musical segments were created from the first two minutes of each piece using Audacity 2.0.4 audio editing software. Then the researcher was assisted to convert the audio files from the windows media audio to the more accessible MP3 format which would enable a greater number of participants to complete the survey across different computers, operating systems and web browsers.

4.4.2.2 Study 1 - B

Qualtrics Survey Software was used to design and host a beta version of the online survey. Each survey item was located on a separate page with forward and backward arrow buttons for participants to navigate between pages. The Qualtrics *Forced Choice* function was attached to all items to ensure that participants completed all items. Attempts to progress through the survey without completing each forced-choice question triggered an error message which would appear in red text at the top of the incomplete page. The message directed the participant back to the incomplete item.

Item 1 of the survey asked the participant to confirm their status as a past or present primary carer for a male with LFA. With the Qualtrics *Skip Logic* function attached, a "no" response to this item deemed the participant ineligible, navigating them automatically to the end of the survey. At the end of the survey, participants were thanked for their participation. *Item 2* enabled the collection of primary carer demographic information such as country, state and area/post code. This item was included for the collection of data that could be statistically analysed for possible associations between demographics and music segment ratings. Responses to *Item 3* asked primary carers to indicate if participants had undertaken formal music training. If the primary carer did not respond in the affirmative to this question, *Skip Logic* automated a transition to *Item 5* as *Item 4* would have not be relevant. An affirmative

response to *Item 3* progressed primary carers to *Item 4*, which asked them to describe the total years of training and type of musical training. *Item 5* provided participants with information regarding the scope of the research and how to operate the electronic survey, confidentiality and ethical details. In addition, participants were informed that they could withdraw at any stage by closing the browser. The contact details of the researcher were also provided for technical or ethical concerns. *Item 6* enabled primary carers to consent to participation with *Skip Logic* attached to skip the primary carer to the end of the survey if consent to participate was not indicated. *Items 7*, *8*, *9*, *10*, *11* and *12* enabled participants to listen to and rate how calming each musical segment would be for the LFA person for whom they cared by operating the Google Audio Player. The code for the audio player was written by a Qualtrics Survey Software representative. Each of the musical segments were presented to participants to rate how calming they would be if listened to by their sons with LFA by using a seven-point Likert-type scale ranging from "*Not at all Calming*" to "*Very Calming*" along with the instructions; “Play, listen then rate!” The word *calming* was substituted for the word *sedate* as published by Hooper et al. (2010) to facilitate a more understandable interpretation. The order of musical segment presentation for these items used the Qualtrics *Randomisation* function to control for order effects and each musical segment was de-identified to avoid musical composer or segment title bias.

Primary carers of males with LFA rated the music due to the researcher’s concerns that the ID component of the LFA diagnosis may affect the ability of those with LFA to comprehend an operational definition of the word *calming* and use that to operate the seven-point Likert-type scale. This premise had been supported by previously published peer-reviewed research (Hooper et al., 2010; Maglione et al., 2012). At the conclusion of the complete survey, participants were thanked for their participation.

Once this beta version of the survey was completed, the survey was analysed for functionality by the researcher in collaboration with the aforementioned Qualtrics Survey Software representative. Initial testing revealed a time delay of ten to 30 seconds between selecting the play button on the Google Audio player and music production. Upon consulting with the representative, such a time delay had not previously occurred as previous Qualtrics surveys had not embedded music. After investigation and further testing, the delay was found to be the result of the music being

held on an external computer server. Therefore, the music was uploaded onto a local server and the time delay was eliminated.

Once the testing had been completed, the researcher published the survey and Qualtrics created a web-link (see Appendix N). The web-link was then attached to an email and sent to national and international autism service providers requesting recruitment opportunities. The service providers were identified via Google search terms such as 'autism', 'autism services' and 'autism services in various countries'. Once participants selected the web-link they were transferred electronically to the beginning of the survey.

Partial and complete survey responses were automatically stored on the Qualtrics Survey Software database. Each respondent was automatically assigned a 10-digit identification number by Qualtrics. No identifying information was indicated by this number. The web-link remained live for a period of four months. At the conclusion of this time, data from all items were downloaded from the Qualtrics Survey Software database to the researcher's Microsoft Excel 2010 program before being analysed using Statistical Package for the Social Sciences (SPSS Version 22). The Qualtrics data were then transferred onto both a password protected computer and Curtin University electronic student account. In addition, hard copies of participant data were printed and stored in a file within a locked cabinet which was located in a secure facility. Access to all electronic and hard copy data was restricted to the researcher and his supervisory committee.

4.5 Data Analysis

A generalised linear mixed model (GLMM) was tested in order to determine whether ratings of calmness varied significantly across the six musical segments. The GLMM was implemented through SPSS's (Version 18) GENLINMIXED procedure. The GLMM represents a special class of regression model. The GLMM is "generalised" in the sense that it can handle dependent variables with non-normal distributions (such as the 7-point "calming scale" used in this study); the GLMM is "mixed" in the sense that it includes both random and fixed effects. For the present GLMM, there was one nominal random effect (participant), one categorical fixed effect (Musical Segment), one binary fixed effect (Formal Music Training: yes, no), and the Musical Segment x Formal Music Training interaction. The Musical Segment fixed effect was a repeated measures factor consisting of six levels.

4.5.1 Statistical assumptions and power

The traditional mixed (consisting of a within-subjects factor and a between-subjects factor) ANOVA model requires the following assumptions to be satisfied: normality, homogeneity of variance, and sphericity. The GLMM "robust statistics" option was invoked to accommodate violations of normality and homogeneity of variance. Violations of sphericity across the within-subjects factor were controlled by changing the covariance matrix from the default of compound symmetry to autoregressive. Also, GLMM is robust to unequal group sizes; in the current design there were 23 participants without but only 9 with formal music training. According to G*Power, the 32 participants provided an 80% chance of detecting a "moderate" (f = 0.19) Musical Segment x Formal Music Training interaction at an alpha-level of .05.

4.6 Results

The means and standard deviations for the Musical Segment x Formal Music Training design are reported in Table 4.3.

Table 4.3

Study 1: Means and Standard Deviations for Musical Segment x Music Training

Formal Music Training	Musical Segment	Mean	*N*	SD
Yes	1	4.33	9	1.22
	2	4.33	9	1.11
	3	3.44	9	1.81
	4	3.67	9	1.93
	5	5.22	9	1.71
	6	5.44	9	2.45
	Total	4.41	54	1.83
No	1	4.78	23	1.44
	2	3.91	23	1.70
	3	3.52	23	1.83
	4	3.74	23	1.32
	5	6.13	23	.92
	6	5.83	23	1.23
	Total	4.65	138	1.74
Total	1	4.66	32	1.38
	2	4.03	32	1.55
	3	3.50	32	1.79
	4	3.72	32	1.48
	5	5.88	32	1.23
	6	5.72	32	1.63
	Total	4.58	192	1.77

Note. 1 = K. 511 (Rondo); 2 = K. 356 (Rondo); 3 = TVK. 265 (Theme and Variations); 4 = BWV 988 (Theme and Variations); 5 = No. 8 Op. 13 (Sonata); 6 = No. 14 Op. 27 (Sonata).

There was a significant main effect for musical segment (F[5,180] = 7.13, p < .001), indicating that segments were not considered to be equally calming. Interestingly, formal musical training did not interact with musical segment (F[5,180] = 1.37, p = .237), indicating that the trend observed in Figure 4.5. applies to those with and without formal music training.

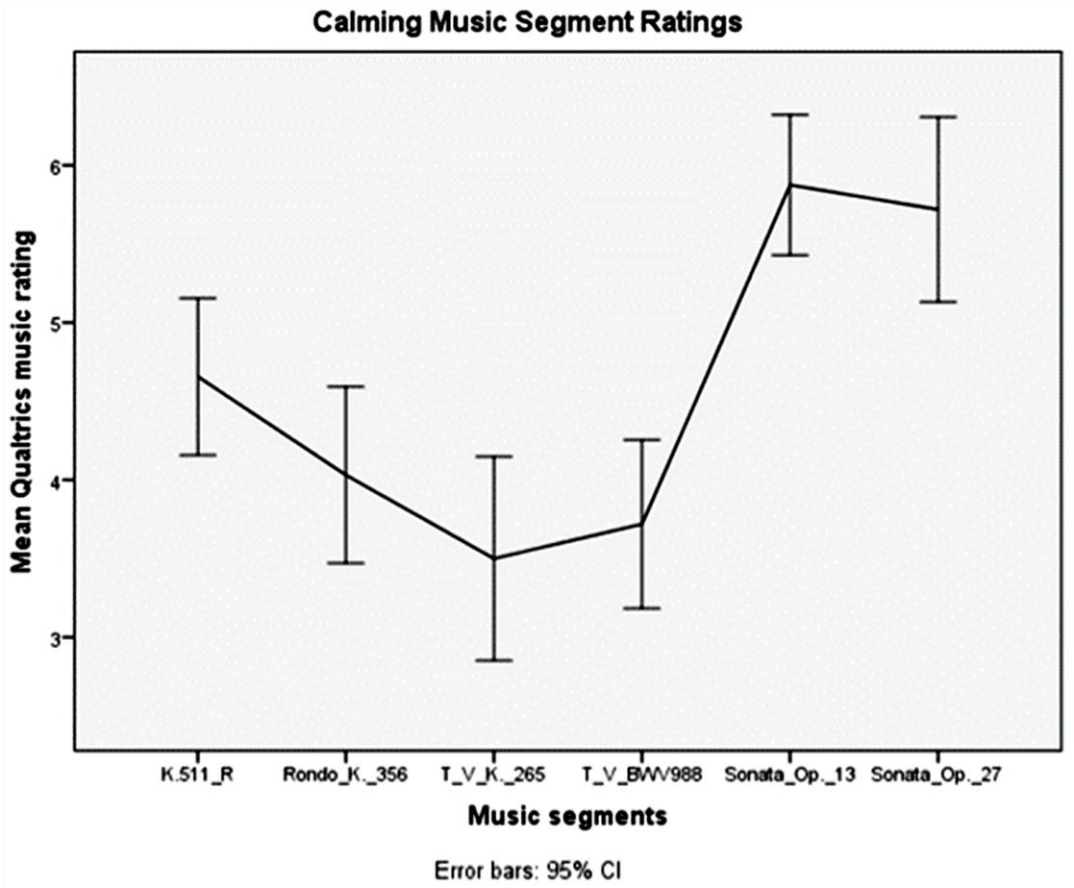


Figure 4.5 Mean music segment ratings with data labels. K.511_R = Wolfgang Amadeus Mozart - Rondo for piano No. 3 in A minor, K. 511; Rondo_K._356 = Wolfgang Amadeus Mozart - Adagio for glass harmonica in C major, K. 356 (K. 617a); T_V_K.265 = Wolfgang Amadeus Mozart - Variations for piano (12) in C major on 'Ah, vous dirai-je maman' K. 265 (K. 300e); T_V_BWV988 = Johann Sebastian Bach's Goldberg Variations, BWV 988-variation 13(2); Sonata _Op._13 = Ludwig van Beethoven's sonata No. 8 in C minor, Op. 13, No. 2. adagio cantabile known as *Sonata Pathetique*; Sonata_Op._27 = Ludwig van Beethoven's sonata no. 14 en do dièse mineur, Op.27, No. 2 - 1 adagio sostenuto known as the *Moonlight Sonata*.

Least Significant Difference (LSD) post-hoc comparisons conducted across the main effect for musical segment indicated that Beethoven's Sonata _Op._13 and Sonata_Op._27 were rated as significantly more calming than Mozart's K.511_R ($p < .001$, $p = .005$), Mozart's Rondo_K._356 ($p < .001$), Mozart's T_V_K.265 ($p < .001$), and Bach's T_V_BWV988 ($p < .001$). Beethoven's Sonata _Op._13 and Sonata_Op._27, were rated as equally calming ($p = .897$) (as seen in Figure 4.5).

4.7 Discussion

The purpose of Study 1 was to design and administer a method of selecting music for music-based interventions. It was hypothesised that a single most calming musical segment could be identified for use in Study 2 and Study 3 of this thesis. Statistical analyses assessing the validity of a GLMM revealed that the six musical segments were not rated as equally calming. In particular, two musical segments composed in sonata form and by Ludwig van Beethoven were found to be significantly more calming than rondo and theme and variations forms composed by Mozart and Bach.

Study 1 employed a music selection method which may be useful for future music-based interventions designed for children with LFA and more generally. This music selection method incorporated expert musician consultations, musical period, form, musical segments and purposeful selection to identify a calming music playlist. In addition, Qualtrics Survey Software was used to build an online survey which reduced the possibilities of selection bias via music de-identification and order effects via randomisation. As a result, Beethoven's Sonata's No. 8 Op. 13 known as the *Sonata Pathetique* and Sonata No. 14 Op. 27 known as the *Moonlight Sonata* were identified as equally calming from the playlist. Given that one musical segment was required for the remaining two studies of the present research, the concert pianist selected the final segment. As result, the *Sonata Pathetique* was chosen to be used to potentially calm school-aged boys with LFA in Study 2 and Study 3.

Incorporating musical form enabled the identification of a musical structure in addition to a composition title. Of interest, as a result of the GLMM analysis, the first 2 minute segments of Beethoven's *Sonata Pathetique* and *Moonlight Sonata* were rated as equally most calming by primary carers. The validity of this finding was promoted by an equal representation of Rondo, Theme and Variations and Sonata musical forms in the calming music playlist. The music ratings, provided by an international sample of 32 primary carers for males with LFA, indicated that music

composed by Beethoven in Sonata form may be most appropriate for calming music-based interventions. Despite this indication, upon listening to the first 2 minutes of Beethoven's *Sonata Pathetique*, it appeared to resemble a rondo musical form (see Figure 4.2). In essence, by reducing the complete sonata to the first 2 minutes, the *Sonata Pathetique* became misrepresented as a rondo. In response to this dilemma, future research may consider selecting longer durations of musical segments. However, this will undoubtedly increase the time required to rate each piece but may help avoid the situation of this study where it was difficult to determine if the rondo or sonata form would be more calming for children with LFA. Further, given that the two musical segments rated as most calming were composed by Ludwig van Beethoven, the identification and analysis of other pieces created by this composer may also be meaningful to this cohort. In addition to the identification of existing music, modern day composers may consider composing music in rondo form for music-based interventions designed to calm children with LFA.

The music selection framework employed by the concert pianist added further rigour to the music selection process by combining evidence-based elements, established musical forms, scientific techniques and musical expertise. As a result, the framework identified a calming music playlist inclusive of six musical segments. The researcher re-assessed the music playlist against its framework prior to distributing the Qualtrics survey to further validate the selected music. Although it is acknowledged that additional musical forms of increased duration may have been preferable, future music-based interventions may give this due consideration.

Cannella et al. (2005) reported that giving people with profound and severe disabilities choice in interventions can result in decreases in undesired behaviour and increases in desired behaviour. Similarly, the present researcher would have preferred it if those with LFA could have rated the calming properties of the musical segments themselves. However, it was believed that the complexity of comprehending and operating a seven-point Likert-type scale and confirming the understanding of the word *calming* may have required cognitive capabilities in excess of what was likely to be possessed by individuals with LFA. Hence, primary carers were included to rate the music. In support, Hooper et al. (2010) reported that primary carers may be appropriate for such tasks.

4.8 Summary

Historically, marketing of the alleged benefits of the *Mozart Effect* without sufficient scientific validation has contributed to music being seen as a treatment of last resort. One of the primary methodological flaws associated with the Rauscher et al. (1993) study, which was the basis of the *Mozart Effect*, was that the authors did not justify why or how this music was selected. Unfortunately, a review of published music-based interventions for children with LFA revealed similar oversights. Without ample scientific rigor used to select music for music-based interventions, the validity, reliability and generalisability of results may remain questionable. Despite some preliminary attempts to select music with more rigour, it is mostly limited.

To the researchers' knowledge, the present study may be one of the first attempts to establish a music selection methodology for music-based interventions designed to treat children with LFA. It is suggested that future music-based interventionists specifically investigate the use of the first two minutes of Beethoven's *Sonata Pathetique* and *Moonlight Sonata* to calm children with LFA. Secondly, it is suggested that music composed by Beethoven be investigated for use in music-based interventions for children with LFA with the intention of calming. Thirdly, it is suggested that researchers consider consulting expert musicians to assist in the selection of music. Finally, future researchers are cautioned when segmenting complete compositions as the form of music can change; as was the case when the first 2-minutes of Beethoven's *Sonata Pathetique* presented in Rondo form once segmented. In sum, it is suggested that the aforementioned method of selecting music may be considered for replication in totality or in part thereof for music-based interventions designed to calm children with LFA. Such replication, in part, may minimise the ongoing negative legacy left by the rise and fall of the *Mozart Effect*. As such, future research may consider employing music selection methods as standard practice for music-based interventions.

CHAPTER 5: Study 2

The aim of Study 2 was to assess the potential effect of music listening on salivary biomarkers of arousal amongst school-aged boys with LFA. Study 2 enabled the assessment of a potential association between two components of the theoretical three-stage mediating model: music listening and arousal (sCort & sAA; see Figure 5.1). A statistically significant relationship between these two components would enable an assessment of the complete model in Study 3. It was hypothesised that the sCort concentration and sAA activity of the music listening group would decrease significantly more than those of the no music listening group after being exposed to a morning school bus ride simulation administered in a controlled environment.

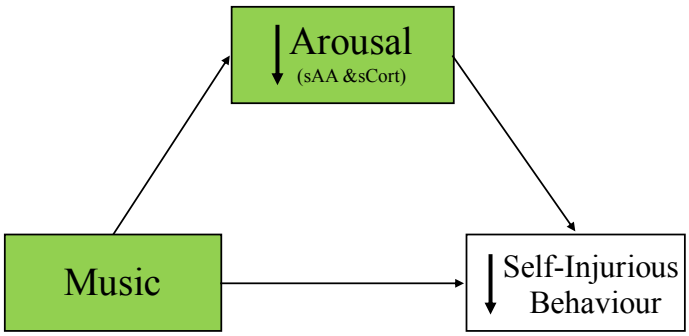


Figure 5.1 Study 2: Three-stage mediating model.

5.1 Method

5.1.1 Participants

Thirty males ranging in age from 4 to 14 years (M = 9.45 years, SD = 3.31 years) participated in Study 2. The participant's LFA status was confirmed by their primary carers as a component of written consent prior to inclusion (see Appendix P). Participants randomly allocated to the music listening group ranged in age from 4 to 14 years (M = 8.93 years, SD = 3.58 years). Participants randomly allocated to the no music listening group ranged in age from 6 to 14 years (M = 9.93 years, SD = 2.89 years). All 30 participants regularly attended government or non-government schools and accessed school bus services for their morning journey to school.

Participants were recruited via telephone and email from government and non-government schools, disability services agencies and parent support groups. The researcher contacted, provided information, and liaised with these schools, agencies and groups directly. A recruitment flyer was distributed to disability services organisations upon request (see Appendix O). The participation of primary carers

involved transporting the participant to the venue which housed the school bus simulator and supporting them to participate. At no stage during Study 2 did primary carers interfere with the research process or protocol. In addition, to activate a snowballing recruitment method, primary carers of participants were asked to tell other potential participants about the research where and when appropriate. This resulted in primary carer recruitment efforts such as social media posts and face-to-face referrals to both schools and individuals.

The inclusion criteria for Study 2 participants were current formal diagnoses of both ASD and an ID consistent with an overall diagnosis of LFA (Cheung et al., 2010). Signed confirmation of this diagnosis was provided by parents/primary carers prior to inclusion (see Appendix P). Participants were excluded from Study 2 if their primary carer believed that they would come to physical and/or psychological harm as a result of being exposed to the morning school bus ride simulator, providing saliva samples or listening to the *Sonata Pathetique*. If this was not predicted, but was believed to be likely during the treatment by the primary carer or the researcher, participation was to be discontinued without prejudice or penalty. As such, informed and signed consent was obtained from the primary carers of participants prior to participation and none of the consenting participants discontinued.

A critical mass of participants was required to identify a possible Music/No Music (Group) x Pre-test/Post-test (Time) interaction via GLMM analysis. This interaction determined whether the music produced a difference over time compared to the no music group. Given the assumptions of a moderate interaction size ($f = .25$), and that the correlation between the pre-test and post-test measures was approximately .6, then 15 participants in each group ($N = 30$) was estimated to yield an 80% chance of detecting the interaction at an alpha-level of .05. According to G*Power, the sample size of 30 provided an 80% chance of detecting a "moderate" ($f = 0.21$) Group x Time interaction at an alpha-level of .05.

5.1.2 Apparatus

5.1.2.1 Saliva sampling and assays

In collaboration with a senior laboratory technician and clinical biochemist, the researcher reviewed several saliva swab and collection tube options which were made available to him within the laboratory. At the conclusion of this process, the researcher, technician and biochemist agreed to use Salimetrics Child Swabs (SCS) and matching collection tubes to sample then store the saliva samples. The SCS was deemed most

appropriate for children with LFA due to its absence of taste, non-abrasive texture and its 12 cm length which minimised choke hazard whilst sampling. As a method of keeping the saliva samples cool between the time of collection and freezing, the researcher used elastic bands to affix the collection tubes to a portable and size-matched freezer block.

Saliva samples were assayed for sCort concentrations using 1-3002 Salimetrics research Cortisol kits (Fortunato et al., 2008). sCort was measured in micrograms per decilitre (µg/dL) of biologically active cortisol concentration in the saliva sample. Samples were assayed for sAA activity with 1-1902 Salimetrics Research Alpha Amylase kits (Fortunato et al., 2008). sAA was measured as units per millilitre of α-amylase activity in the saliva sample. All saliva assay kits were stored in a cool room which maintained a temperature of between 2 and 8 degrees Celsius.

5.1.2.2 Morning school bus ride simulator

The construction and function of the morning school bus ride simulator was made up of numerous elements. Firstly, video footage and audio recordings were required to produce the sights and sounds associated with a naturalistic morning school bus ride. Initially, preliminary testing of a video camera with a boom microphone attached and Sony surround sound audio recorder were undertaken. The researcher and an audio-visual consultant from Curtin University completed this testing on a university shuttle bus service. The intention of this testing was to ensure that the equipment selected for recording video and sound on an actual morning school bus ride was of high enough quality to be used in the simulator. As such, adjustments to the settings of this equipment were crucial to replicate the morning school bus ride with accuracy and realism. At the conclusion of this equipment testing phase, the researcher and consultant agreed that the aforementioned Sony camcorder and surround sound audio recorder were appropriate. A lens was attached to the camcorder to ensure that the recorded field of view was appropriate.

The researcher then contacted the Public Transport Authority of Western Australia–School Bus Services (PTA-SBS) to obtain the contact details of a school bus driver who may allow filming and sound recording to occur on their bus. Subsequently, the researcher attended morning school bus rides and recorded video and audio of the final 15 minutes before arriving at school.

The Sony camcorder was mounted on a tripod that had been affixed to the floor of the school bus at the base of a seat located in the first row. The surround sound audio

recorder was mounted on a separate tripod to the left of the camcorder. A black-coloured adhesive square was affixed to the inside of the front bus windscreen to enable the measurement of the recording location (see Figure 5.2). These measurements were used to ensure that the location of the school bus seat in the simulator matched the location of the bus seat on the actual school bus from which the video and audio recordings were collected.

Figure 5.2 Study 2: Screen image of the morning school bus simulator field of view featuring the black-coloured adhesive square.

After the fourth recording session, the researcher had captured the video and audio which was of quality to be used in the simulator. In addition, the researcher believed that footage recorded on the final week would be a more accurate representation of a usual morning bus ride as the students on the bus would have been desensitised to the presence of the researcher by this stage.

The school bus simulator was constructed and housed in a secure and controlled facility on the campus at Curtin University. The researcher was granted access to a white-walled, carpeted, windowless 7.6m long and 3.05m wide room within which to construct the simulator. The internal fittings of the room included a ceiling mounted projector, 2.92 m height and 2.04 m width wall mounted projector screen, school bus seat pair with seat belts mounted on an aluminium base affixed to carpet by hook Velcro, 5.1 surround sound system, audio-visual control desk with computer, primary carer observation seat and 1.2m wide entry door. The aluminium

base of the school bus seat pair was manufactured by a metalwork fabricator to the exact specifications of a design created by an engineer to minimise tip risk for participants (see Figure 5.3). Tip risk was also minimised by affixing the aluminium base to the carpet on the floor of the room using the hook side of strips of industrial strength Velcro. The seat belt attached to the inactive seat of the school bus seat pair was removed to ensure that the participant sat in the designated seat only. All materials for the internal fittings were donated by the Properties Department of Curtin University, local audio-visual retailers and school bus manufacturers.

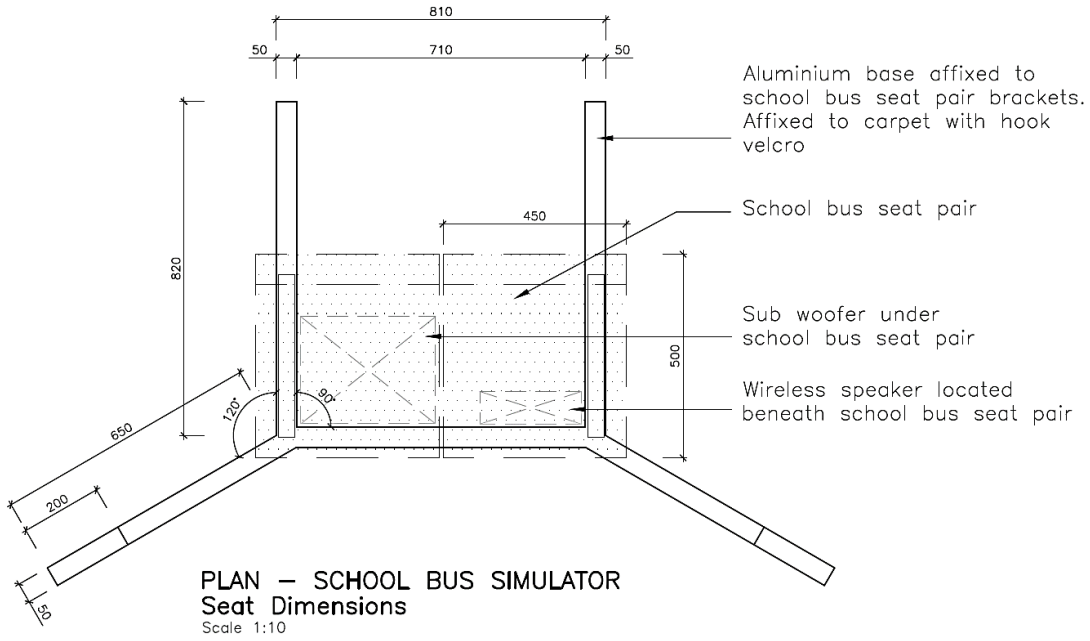


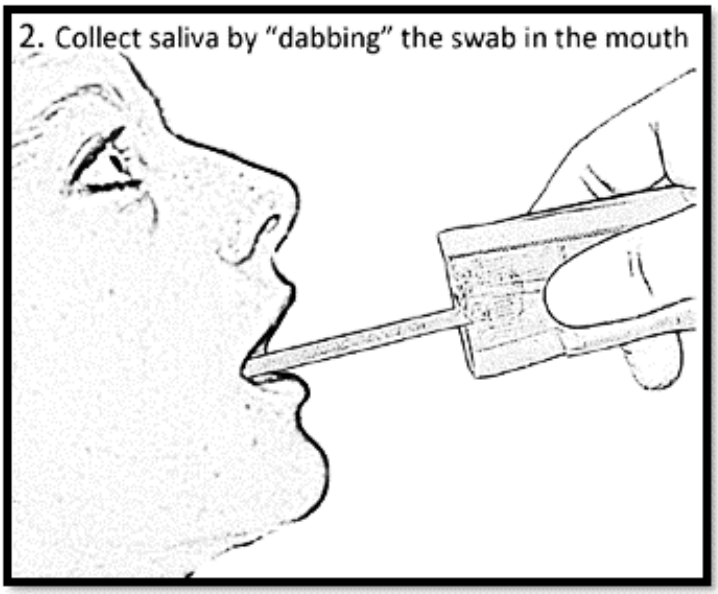
Figure 5.3 Study 2: To scale diagram with specifications of the school bus seat pair.

The video footage recorded on the bus was reduced to a duration of 14 minutes and 35 seconds to match the duration of the seven repeats of the first 2 minutes and 5 seconds of Beethoven's *Sonata Pathetique*. This ensured that it exceeded the 13-minute demand condition presented in the original TSST research as published by Kirschbaum et al. (1993). The video footage recorded on the final day of the recording schedule included children with disabilities arriving at school and being dropped off by primary carers, the appearance of an unidentifiable school bus driver driving the bus, cloudy skies with dry road conditions, and various features of suburban streets before an arrival at a school for children with special needs. The footage indicated this arrival when a bus in front was seen to assist two students depart in wheelchairs using a wheelchair hoist. In addition, the audio component of the recording featured a student spontaneously verbalising "teachers, teachers teachers" as the school bus came to a

rest. It is suggested that these features may have provided participants with a clear indication that they had arrived at a school for children with special needs, a scenario that the participants with LFA would have become accustomed to. The audio recording was reduced to a duration that matched the video footage before being combined. The final combined audio and visual for the morning school bus ride simulation was then saved as a 2.18GB MP4 formatted video file.

The researcher assumed that the participants may not tolerate wearing earphones as a result of likely "hyper- or hyporeactivity to sensory input" and "adverse response to specific sounds" associated with the ASD component of their LFA diagnosis (American Psychiatric Association, 2013, p. 51). Such intolerance had been reported previously by Hooper et al. (2012). Therefore, the researcher investigated the functionality of numerous types of wireless Bluetooth enabled speakers. A range of speakers were then tested by the researcher for sound volume and clarity both in the researcher's car and on the Curtin University student bus. As a result, a BIG Jambox brand wireless speaker manufactured by a company named *Jawbone* was chosen. The music was transferred via Bluetooth technology from a tablet computer to the wireless speaker located beneath the school bus seat of the participant.

Upon acquiring the apparatus and audio-visual materials required to complete the school bus simulator, all components were assembled. Firstly, the position of the seat, as per the calculations recorded on the actual school bus, was fixed to the carpet by the Velcro. The 5.1 surround sound system was then connected to the control desk computer. White-coloured adhesive tape was affixed to the floor to indicate to the participant and their primary carer where they were required to walk once entering the simulator. Several tubes of the overhead fluorescent lighting within the simulator were deactivated to approximate the level of light detected by the researcher on the day of recording. Similarly, testing was undertaken to ensure that the volume of noise produced by the surround sound system approximated that of the actual school. A locked cabinet was used to store the saliva swabs and collection tubes. All electrical cords associated with the surround sound system were then affixed to the carpet with black-coloured adhesive tape, the primary carer saliva sampling instruction was affixed to the wall and the school bus simulator set-up was complete (see Figures 5.4 & 5.5).



20-30 seconds under the front of the tongue

Figure 5.4 Study 2: Saliva collection instructions for primary carers.

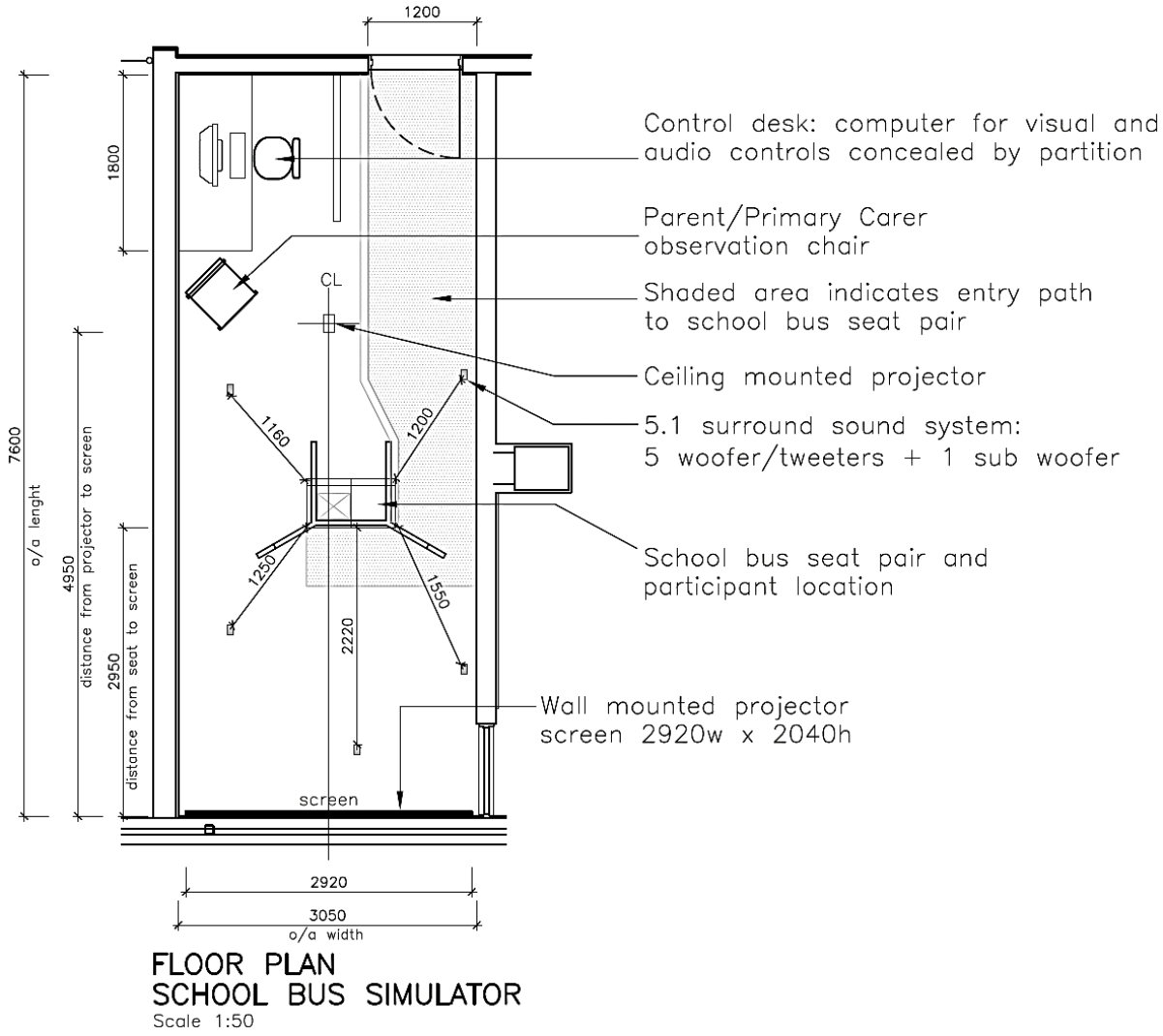


Figure 5.5 Study 2: School bus simulator photograph and to scale diagram with specifications.

After the simulator construction was complete, the researcher and audio-visual consultant tested the simulation. Whilst sitting in the assigned participant school bus seat, the researcher began to feel slightly unwell; a phenomena known as simulator sickness. For some, simulator sickness can elicit dizzy, faint or ill sensations whilst in a simulator (Stanney & Salvendy, 1998). Reason and Brand (1975), as cited in Brooks et al. (2010), suggested that these sensations can be the result of sensory conflicts, such as visual and vestibular, as the body attempts to process the virtual environment. A review of the literature conducted by the researcher revealed that the best method of avoiding such sensations was to ensure that the simulator was as realistic as possible. Therefore, an ultraviolet lens was removed from the overhead film projector to ensure that the footage was of highest clarity. None of the participants appeared to experience

symptoms of SS, engaged meaningfully with the simulation and ate a breakfast as provided by the researcher after participating.

5.2 Procedure

As per Study 1, the Curtin University Human Research Ethics Committee assigned the approval number of HR138/2011 also pertained to this study. In addition, ethical approval was sought and obtained via the Western Australian Department of Education and Training, the Centre for Cerebral Palsy of Western Australia, the Western Australian Autism Registry and the Public Transport Authority of Western Australia–School Bus Services ethics committees. Information letters for Study 2 were distributed and signed consent was obtained from the primary carers of participants prior to the commencement of this study (see Appendix P).

Throughout the study, the ethical guidelines relating to research involving children was upheld as stipulated by the *National Statement on Ethical Conduct in Humans Research* published by the National Health and Medical Research Council. In addition, registration as an authorised clinical trial was obtained from the Australian New Zealand Clinical Trials Registry (ANZCTR) and the Western Australian Register for Autism Spectrum Disorders. The ANZCTR provided the authorisation number of 12611001063909. This authorisation number appeared on all related electronic and hard copy information regarding Study 2.

The physical and psychological wellbeing of the participants was of paramount importance throughout this study. The researcher had accumulated in excess of 20 years' prior experience working directly with children with LFA and their primary carers. As such, the researcher administered all aspects of the research. No treatments were discontinued for any reason throughout the research nor were any concerns raised directly by primary carers to the researcher or the Curtin University Human Research Ethics Committee. To maintain confidentiality and anonymity, participants and their saliva samples were identified via an individualised 3-digit code (Tate & Baroff, 1966; Wachtel et al., 2009; Wachtel et al., 2011).

Primary carers consenting to their children's participation in Study 2 were provided with an information letter (See Appendix Q). This letter contained essential information regarding the study such as date, time, and requirements such as nil by mouth and to not brush or floss teeth on the morning of the treatment. The primary carer was also informed that breakfast would be provided at a local cafe by the researcher after the treatment had concluded. Lastly, the primary carer was informed

that he/she would be required to collect the saliva samples as instructed, assisted by the researcher. The letter also contained what is known as a *Social Story* (Gray & White, 2002). This pictorial story can be used to provide meaning and a specific description to situations such as the trip to the school bus simulator for children with disabilities such as those with LFA participating in Study 2 (see Appendix Q). The researcher hypothesised that the story may help the participants cope with likely "inflexible adherence to routines" and "need to take same route" associated with the ASD component of their LFA diagnosis (American Psychiatric Association, 2013, p. 50). This information letter was sent to the primary carer of participants via email prior to the morning of participation. A telephone call from the researcher to the primary carer of the participant was made prior to the treatment to answer questions and provide a reminder with reference to the content of the information letter. Primary carers were verbally informed by the researcher that confidentially and anonymity would be maintained and that withdrawal from the research at any stage without prejudice or penalty would be supported. At this stage, primary carers were encouraged by the researcher to ask for clarification at any stage prior to, during or after the research had been consented to and/or concluded.

The researcher then underwent biological laboratory training, and obtained an occupational health and safety clearance. Further, the researcher was taught how to assay saliva samples for biomarkers of arousal by the senior laboratory technician under the supervision of the clinical biochemist. The saliva samples and material used in training were not related to the present study. Rather the researcher took samples from colleagues and himself. sCort and sAA outputs were analysed by the clinical biochemist for accuracy and validity prior to the researcher being approved to independently conduct the analyses using participant saliva samples for Study 2. Ongoing supervision was provided in the laboratory by the senior laboratory technician and results reviewed by the clinical biochemist.

Prior to the commencement of the research, the researcher completed the saliva samples' labels in triplicate. The first label was affixed to the saliva swab collection tube to ensure that the sample was identifiable. The second identical label was placed onto a chart which was affixed to the rear of the partition within the simulator venue. The third identical label was affixed to the rear of the original completed hard copy consent form for each participant. All labels were pre-populated with information pertaining to the date of sample, the Curtin University Human Research Ethics

Committee assigned approval number of HR138/2011, patient initials, a sample 3-digit identifier code from 001 to 150 and a sample time-point description of Pre, Post_0, Post_5, Post_10, or Post_20. The simulator was locked to ensure that all information contained within the room remained confidential.

A random allocation sequence was then established to negate the potential confound of order effects. In this pretest-posttest control group design with random allocation of subjects to groups, the participants were *randomly* allocated to either the experimental group (music-listening music) or control group (no music listening). The random allocation sequence was established before the commencement of the first morning school bus ride simulation using the Quickcalcs on-line randomiser application as published by Graphpad Software (2015). The order by which the boys with LFA participated in the trial was stipulated by the random allocation sequence specified in table 5.1.

Table 5.1

Study 2: Participant Random Allocation Sequence by Graphpad Software (2015)

Participant	Condition	Participant	Condition
1	No Music Listening	16	No Music Listening
2	No Music Listening	17	Music Listening
3	Music Listening	18	Music Listening
4	Music Listening	19	Music Listening
5	No Music Listening	20	Music Listening
6	Music Listening	21	No Music Listening
7	Music Listening	22	Music Listening
8	No Music Listening	23	No Music Listening
9	No Music Listening	24	Music Listening
10	No Music Listening	25	Music Listening
11	Music Listening	26	Music Listening
12	No Music Listening	27	Music Listening
13	No Music Listening	28	No Music Listening
14	Music Listening	29	No Music Listening
15	No Music Listening	30	No Music Listening

As seen in Figure 5.6, the design of Study 2 saw participants assigned to control (no music listening) and experimental (music listening) groups in a pretest-posttest control group between-subjects RCT design with random allocation of participants to groups. All morning school bus ride simulations commenced at 0830hrs to match the approximate time of the participants' school bus rides.

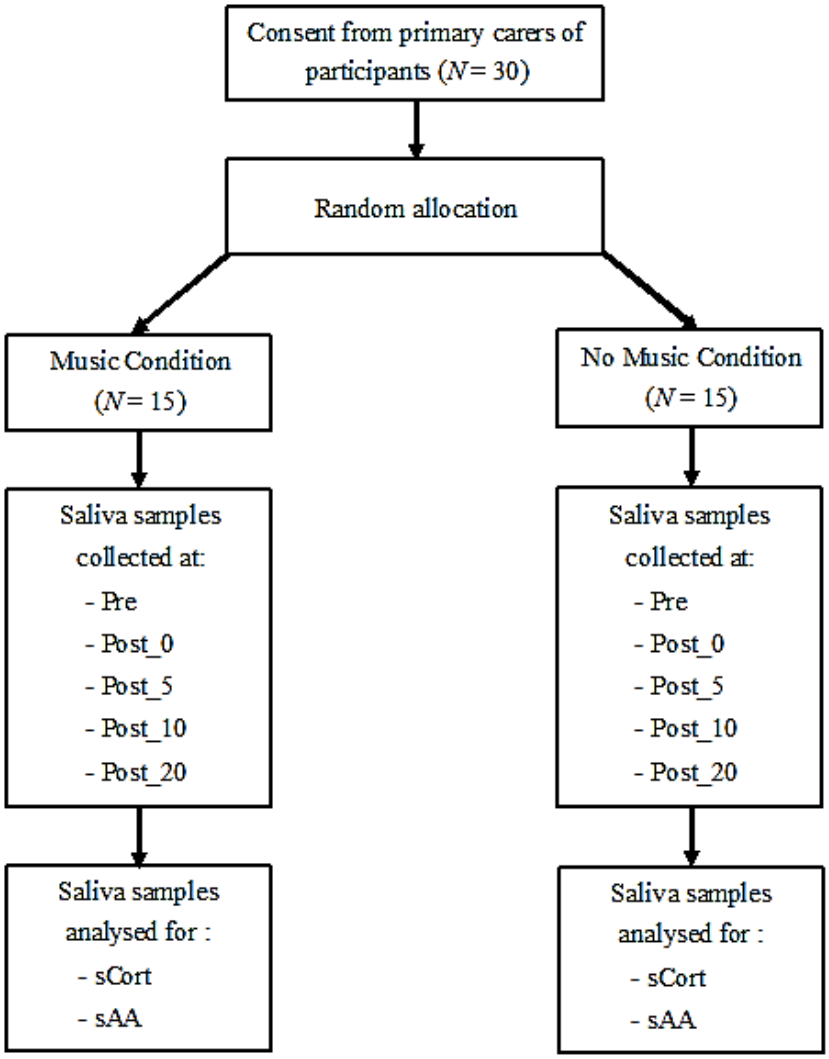


Figure 5.6 Study 2: Flow diagram of the pretest-posttest control group between-subjects RCT design.

On the morning of the treatment, the researcher met each participant and their primary carer in the car park of the simulator venue. The researcher then accompanied the party to the simulator. The participant and primary carer were required to arrive at the simulator venue by 08:15 hrs on the morning of the treatment. This enabled a period of 15 minutes for the researcher to meet the participant, answer any questions posed by the primary carer, have the hard copy consent form signed and ensure that

the participant was seated and ready for the pre saliva sample collection prior to the commencement of the morning school bus ride simulation.

Often, the primary carer used the social story provided in the information letter to remind the participant of the novel environment to which they were about to be exposed. As the participant and primary carer came closer to the simulator, they were able to look inside the simulator room as the researcher purposefully left the door ajar. The researcher often referred to the final image of the social story at this stage as it matched the image seen through the ajar door (see Appendix Q).

The child was then invited by the researcher to enter the simulator and directed to sit in the allocated school bus seat, put on the seat belt and wait for the bus ride to begin after providing a sample of their saliva. At this stage, the door to the simulator room was closed and the primary carer was verbally instructed on the saliva collection protocol and provided signed consent. Given that the primary carer had previously been emailed an information letter with the social story, few questions were posed to the researcher at this stage.

The pre-saliva sample was then collected by the primary carer immediately before 8:30am and the simulator was activated. The protocol for the collection of each saliva sample was as follows. The researcher unwrapped the SCS, offering the dry end to the primary carer. The primary carer then took the swab and collected the sample from the mouth of the participant for a period of 30 seconds as recommended in the SCS instructions and prompted by the instruction sheet which had been affixed to the wall (see Figure 5.4). The dry end of the SCS was then provided to the researcher for assessment. The researcher assessed each SCS for saliva yield via visual inspection and hand weighing. If the SCS was deemed by the researcher to contain ample saliva for sCort 25µL and sAA 15µL assays, it was placed in the pre-labelled collection tube before being affixed to the portable freezer block using 2 elastic bands. If the SCS did not appear to yield an ample sample, the researcher requested that the primary carer to re-apply for an additional 30 seconds. On four occasions where this occurred, the SCS was found to contain an ample sample after the second administration.

For the participants randomly allocated to the music listening condition, the researcher manually activated the wireless speaker in synchrony with the commencement of the simulation. At the conclusion of the simulation, 14 minutes and 35 seconds, the Post_0 sample was collected by the primary carer being supervised and assisted by the researcher. At this stage, the researcher activated a stop watch to

ensure that the final three saliva samples were collected in a timely manner. Hence, the primary carer was supported by the researcher to collect the Post_5, Post_10 and Post_20 saliva samples at the appropriate times. Between the Post_0 and Post_20 saliva samples, the participant was allowed to engage in free play in the research venue. At the conclusion of the Post_20 sample, the participant and primary carer were accompanied by the researcher to a café located in the building and provided with a breakfast of their choice. During breakfast, the researcher employed the snowballing recruitment method and asked for verbal feedback about the research experience. At the conclusion of breakfast, the participant and primary carer were accompanied by the researcher back to the carpark, thanked for their participation and departed the venue.

The saliva samples were then conveyed immediately to the Curtin University biomedical laboratory by the researcher before being centrifuged at 3000 rpm and pipetted from the collection tube into pre-labelled storage tubes. The storage tubes were labelled with participant initials, a saliva sample 3-digit identifier code from 001 to 150 and a sample time-point description (Pre, Post_0, Post_5, Post_10, and Post_20). The samples were then placed in the freezer and subsequently frozen within 60 minutes of the collection of the Post_20 sample. Once all 150 samples for the study had been collected, they were thawed and assayed in duplicate for sCort concentrations and sAA activity.

5.2.1 Saliva assays

One sCort assay run, populating a 96-well microtitre plate to analyse 38 samples in duplicate, took a total of five hours to complete. Plate plans were completed on A4 template paper and endorsed by the clinical biochemist with senior laboratory technician prior to assaying. Five sCort assay plates were completed and validated for precision by the clinical biochemist and senior laboratory technician. The absorbances of raw samples were read via photometry at 450 nanometers (nm) per 0.1 second in a Victor Fluoro plate reader set to room temperature. The plate reader was linked to a password protected computer running the Windows 7 operating system installed with the Wallac 1420 Version 3.0 computer software manufactured by Perkin Elmer Life Sciences. At the conclusion of each assay run, Wallac 1420 automatically stored the raw data into a 2010 Microsoft Excel spreadsheet. This spreadsheet was then transferred to the researcher's password protected computer and saved with a title representing the biomarker and sample identifications it contained. The raw sCort

concentration absorbance data were then uploaded into the on-line sCort program of MyAssays: Analysis Software Solutions (2015) where it underwent a Four Parameter Logistic Fit transformation into micrograms per decilitre (µg/dL) of biologically active cortisol concentration contained in each duplicate saliva sample. These data were then transferred to a password protected 2010 Microsoft Excel spreadsheet and saved by title and sample identification as the final data set. Additional data recorded in the final Excel spreadsheet included the raw data, covariates, precision plots, average concentrations, standard deviations, standard errors and a photograph of the plate plan used.

The analysis of saliva samples for sAA activity was based on measurements of its enzymatic activity using a chromogenic substrate. Assays were run in 96-well microtitre plate frames, populating only 40-wells to analyse 18 samples in duplicate, with each plate taking the researcher a total of three hours to complete. The remaining 56 wells were removed, stored and re-used as needed. A total of 12 sAA assay runs were completed and validated by the clinical biochemist and senior laboratory technician. The same Victor Fluoro plate reader linked to a computer running the Windows 7 operating system with Wallac 1420 Version 3.0 computer software was used to analyse the sAA data. The plate reader was specifically programmed to read enzymatic absorbances at 450 nanometers (nm) per 2 minute increments at 37^{o}C, for a total of 8 minutes. Linearity of the enzyme rate was checked by linear reaction (based on a plot of absorbance vs time) and converted to reaction rate and expressed as Units/mL. All sAA data files underwent the same password protection, title labelling tab inclusions as the sCort files.

Given that all samples were analysed in duplicate, intra-assay precision could be determined on the basis of all samples analysed, and expressed as a coefficient of variation (CV = SD/mean x 100). Samples with a CV in excess of 10% were re-assayed. Such repeat analyses invariably yielded a lower CV. The sCort concentration ranged from approximately 0.01 µg/dL to approximately 1.7 µg/dL, with the intra-assay precision ranging from 5% to 2% over that concentration range. sAA activity ranged from approximately 15 U/mL to approximately 250 U/mL. The intra-assay assay precision was excellent, averaging 2.0%, and ranged from 1% at 80 U/mL to 2% at 250 U/mL.

5.3 Data Analysis

A GLMM, implemented through SPSS's (Version 22) GENLINMIXED procedure, was used to test whether time-related changes in sCort concentrations and sAA activity varied significantly between music listening and no music listening conditions. The GLMM is "generalised" in the sense that it can accommodate dependent variables with non-normal distributions (such as the 7-point "calming scale" used in Study 1, and the sCort concentration and sAA activity in this study). For the present GLMM, there was one nominal random effect (participant), one ordinal fixed effect (time), one binary fixed effect (group: music listening, no music listening), and the time x group interaction. The time fixed effect was a repeated measures factor consisting of five levels: Pre-TSST-C, Post_0, Post_5, Post_10, and Post_20). To optimise the likelihood of convergence, separate GLMM analyses were run for the sCort and sAA data.

5.3.1 Statistical assumptions

As with the analysis of data from Study 1, the GLMM "robust statistics" option was invoked to accommodate violations of normality and homogeneity of variance; and violations of sphericity across the within-subjects factor were controlled by changing the covariance matrix from the default of compound symmetry to autoregressive.

5.4 Results

The sCort concentration means and standard deviation for the Group x Time design are reported in Table 5.2.

Table 5.2

Study 2: sCort Concentration Means and Standard Deviations for the Group x Time Design

Group	Time	*N*	Mean	Standard Deviation
No Music	1	15	.26	.17
	2	15	.22	.16
	3	15	.20	.14
	4	15	.23	.26
	5	14	.21	.16
	Total	74	.22	.18
Music	1	15	.30	.26
	2	14	.23	.16
	3	15	.23	.14
	4	14	.18	.11
	5	15	.18	.08
	Total	73	.22	.17
Total	1	30	.28	.22
	2	29	.23	.16
	3	30	.21	.14
	4	29	.21	.20
	5	29	.19	.12
	Total	147	.22	.17

For sCort, there was a significant main effect for time ($F[4,137] = 3.35$, $\eta^2_p = .022$, $p = .012$) but no significant effect main for group ($F[1,137] = 0.00$, $\eta^2_p = .00$, $p = .959$). Group interacted significantly with time ($F[4,137] = 2.79$, $\eta^2_p = .012$, $p = .040$), indicating that time-related changes in sCort concentrations varied across the two groups. Figure 5.8 plots the interaction. As seen in this figure, the interaction at the Post_5 to Post_10 saliva sample time-points section of the sCort concentration trajectory show no change for the no music listening group ($p = .395$) but a significant decrease for the music listening group ($p < .001$).

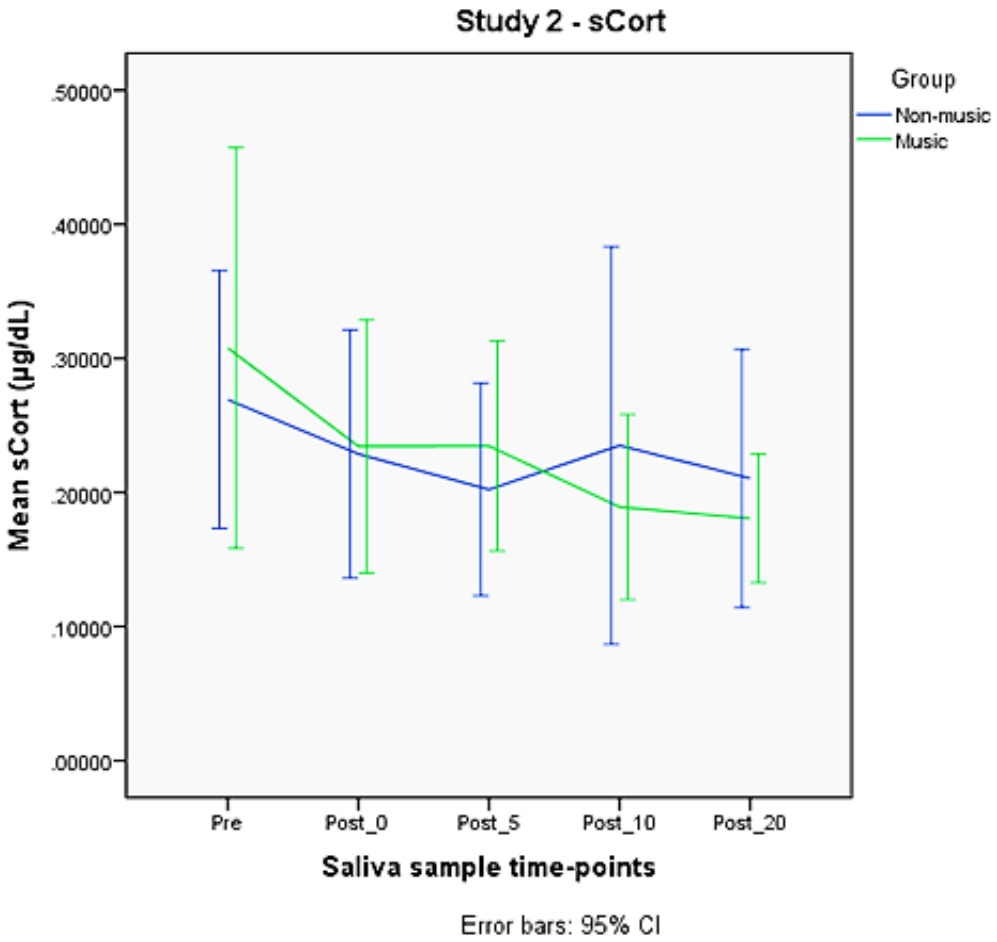


Figure 5.7 Study 2: Mean salivary cortisol (sCort) concentrations in micrograms per decilitre (µg/dL) of the music listening and no music listening groups.

The sAA activity means and standard deviation for the Group x Time design are reported in Table 5.3.

Table 5.3

Study 2: sAA Activity Means and Standard Deviations for the Group x Time Design

Group	Time	*N*	Mean	Standard Deviation
No Music	1	15	135.47	110.08
	2	15	132.56	86.51
	3	15	143.69	108.70
	4	15	125.80	70.77
	5	15	121.56	66.20
	Total	75	131.82	88.21
Music	1	15	148.43	163.05
	2	15	128.56	121.74
	3	14	122.15	122.57
	4	15	101.78	87.85
	5	13	108.33	102.72
	Total	72	122.22	120.41
Total	1	30	141.95	136.85
	2	30	130.56	103.79
	3	29	133.29	114.03
	4	30	113.79	79.33
	5	28	115.42	83.71
	Total	147	127.12	104.96

There were no significant main effects for Time (F[4,137] = 1.08, η^2_p = .001, p = .371), no significant effect main for Group (F[1,137] = 0.10, η^2_p = .000, p = .747), and no significant Group x Time interaction (F[4,137] = 0.78, η^2_p = .001, p = .540). The age of the participants was not related to sAA activity. The non-significant effects are plotted in Figure 5.9. Although the Group x Time interaction was non-significant, least significant difference (LSD) contrasts conducted across the simple main effects of time indicated significant decreases in the sAA activity of the music listening group at Pre to Post_10 (p = .027), Pre to Post_20 (p = .026), and Post_0 to Post_10 (p = .044). However, the LSD contrasts across the simple main effect of time for the no music listening group were all non-significant. It should be emphasised, however, that in the presence of the non-significant interaction, the significant LSD contrasts are merely suggestive of effects that might emerge with a larger sample size.

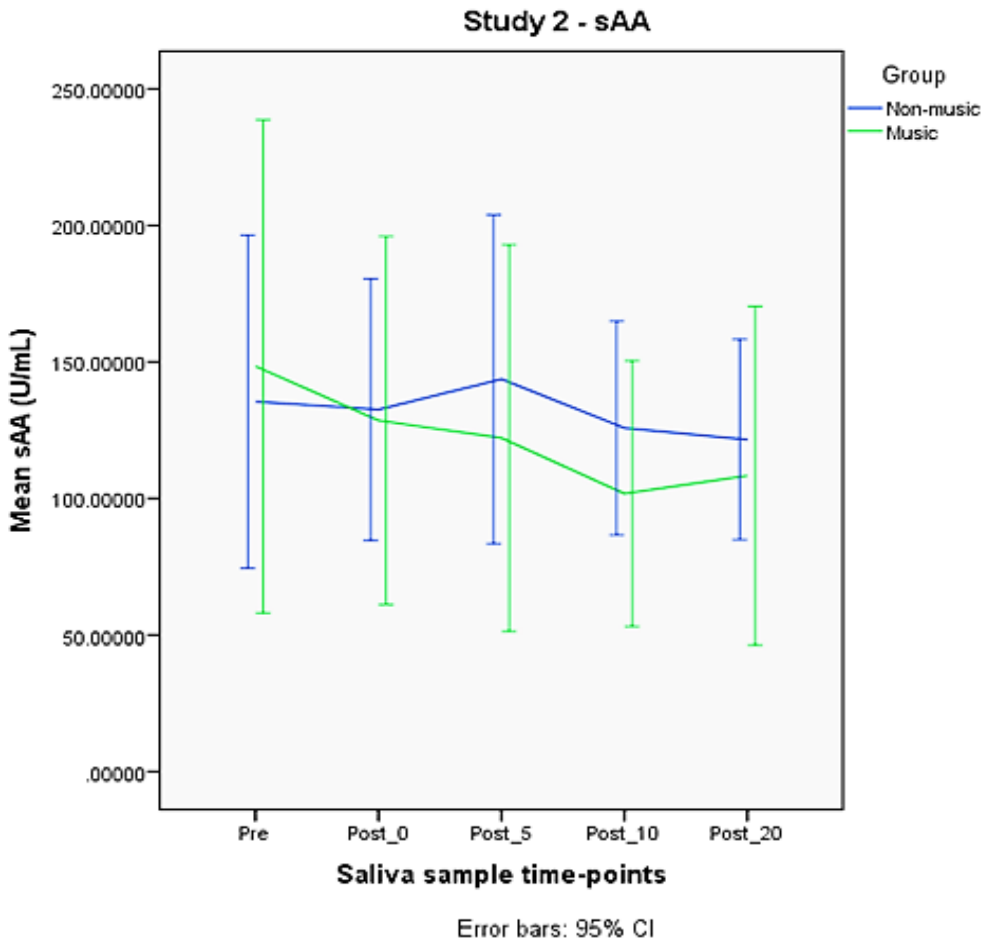


Figure 5.8 Study 2: Mean salivary alpha-amylase (sAA) activity in units per microliter (U/mL) of the music listening and no music listening groups.

5.5 Discussion

The purpose of Study 2 was to investigate the potential for music listening to reduce salivary biomarkers of arousal amongst school-aged boys with LFA before and after being exposed to a demand condition in the form of a TSST-C morning school bus ride simulation. The simulation was designed to pose a safe and realistic laboratory-based demand on the participants and a method of assessing a potential link between the music listening and arousal (sCort & sAA) components of the three-stage mediating model (see Figure 5.1).

As seen in Figure 5.8, a significant sCort concentration decrease amongst the music listening group was detected from Post_5 to Post_10 saliva collection time-points ($p < .001$) when compared to the no music listening group ($p = .395$). Of interest, White and Mulligan (2009) reported that on average, sCort concentration peaks approximately 20 minutes after being exposed to a demand condition. Based on the

above sCort finding, it appears that the significant decrease in activity was detected before the suggested peak. In addition, it has also been reported that changes in sCort concentrations are often most noticeable at either 10 or 20 minutes after exposure to a stressor (Corbett et al., 2012; Quas, 2011). It may be assumed that the significant sCort concentration decrease may have occurred at the lower limit of this range. However, it must be noted that the above reports were based on either an undefined general population (White & Mulligan, 2009), an undefined general population of children (Quas, 2011) or children with ASD but not an ID (Corbett et al., 2012). Hence, their relevance to the significant sCort concentration decrease found in Study 2 is questionable.

People with ASD do not often respond typically to stress (Kidd et al., 2012). Further, Bitsika et al. (2015) suggested that future research focus on assessing the legitimacy of cortisol as an arousal biomarker for children with ASD. With a real-life application, the sCort result may be relevant to primary carers of boys with LFA who are interested in reducing biological arousal for their children when exposed to a demand condition within other controlled environments such as the doctor's rooms, dental surgery and so forth. In the researcher's experience, these environments can be highly arousing for children with LFA. Moreover, it is suggested that future research consider replicating the pre-test and post-test TSST-C design of Study 2 within these demanding environments.

There were no significant main effects nor was a significant sAA activity and time interaction detected. Despite this, the LSD contrasts conducted across the simple main effects of time indicated that the activity of sAA decreased significantly amongst the music listening group from Pre to Post_10 saliva collection time-points whilst the LSD contrasts for the no music listening group on time was non-significant. Despite the sAA results being suggestive at best, it is possible that larger effects could emerge with a larger sample size. Furthermore, the existing scientific body of knowledge regarding ASD and sAA is small (Bosch et al., 2011; Kidd et al., 2012). For this reason, the current sAA results identify an area of possible future research regarding ASD and sAA.

In combination, the results from Study 2 may provide information that is useful when considering the assessment of sCort concentrations and sAA activity amongst boys with LFA in relation to exposure to a demand conditions in controlled environments. Moreover, the results support the hypothesised relationship between the

music listening and arousal (sCort & sAA) posed as components of the three-stage mediating model.

The validity of the sAA and sCort data is potentially strengthened by standardising the saliva sampling time-points. It had previously been reported that standardising the time-points for measuring cortisol could be considered best practice in biomarker research (Bauer et al., 2002). Hence, the time of day chosen for the commencement of the morning school bus ride simulation was standardised to replicate an actual morning school bus ride for participants. However, it was not deemed appropriate to standardise the activity partaken by participants between the Post_0 and Post_20 saliva collection time-points as they would naturalistically partake in many and varied activities upon arrival at school.

Within-subjects experimental designs are often used to detect treatment effects as the participant functions as their own control. However, such designs require individual participants to re-visit treatment venues on numerous treatment days. In the case of the present study, it is suggested that such designs may have resulted in high rates of attrition for numerous reasons. Firstly, as a result of the ASD component of their LFA diagnosis, the participants were likely to not favour frequent changes to routine (American Psychiatric Association, 2013). Further, participation required not only being exposed to the morning school bus simulator demand condition, but not eating breakfast, brushing or flossing teeth on the morning of the trial. Such changes in routine are often seen as untenable and potentially harmful to children with ASD and their families. In addition, within-subject designs are prone to carry-over effects, which may or may not be effectively controlled by counter-balancing (Howell, 2014). It was for these reasons that the pretest-posttest control group design with random allocation of subjects to groups was deemed most appropriate.

The best indication as to the realism of the school bus simulation or the emergence of simulator sickness would have been participant behaviour. However, similar to the point raised in Study 1, it is suggested that information from the boys in relation to these topics may have not been accurate nor readily accessible as a result of the ID component of their LFA diagnosis. Further, providing verbal feedback as to the realism of the simulator may not have been accurate. However, similar to Study 1, the researcher may have asked the primary carers to rate the realism of the simulator based on their knowledge of the actual school bus regularly used by the participant. This may

have been most valid as the primary carer was present for the entire simulation However, this was not employed in Study 2.

Vining and McGinley (1986) suggested that saliva collection techniques such as using a syringe fitted with a plastic tube to exhume saliva from the floor of the mouth or using cotton wool rolls was not advisable as the large surface area of these materials increases the likelihood of non-specific adsorption to confound results. However, the Salimetrics Child Swab (SCS) product used to collect saliva samples for the present research was tested and deemed appropriate for use in Study 2 by the researcher, senior laboratory technician and clinical biochemist for several reasons. Firstly, the 12 cm length of the swab enabled the collection of saliva with minimal choke risk to the participant as the collector was able to hold securely onto a dry end of ample length. The length of the swab also protected the collector from potential bite injuries. Secondly, the high absorbency capability of the SCS ensured that the total time required for saliva sample collection was minimal. As a result, the saliva sampling time-point schedule was maintained. The sampling appeared to be more readily accepted by the participants as the length of time required to endure the collection process was reduced. For example, the average yield of the SCS was reported by Salimetrics to range from 100 µL to 500 µL per sample. This was deemed as more than ample as the total volumes of sample required for sCort was 25 µL and sAA was 10 µL. Lastly, the robustness of the SCS enabled for the effective collection of saliva despite participants biting, chewing and attempting to tear the swabs. Moreover, use of the SCS appeared to enable the collection of saliva from a sometimes challenging cohort of children and yielded adequate sample volumes for assaying.

The validity of the sCort and sAA data and subsequent results was also strengthened by the involvement of the senior laboratory technician and clinical biochemist. The senior laboratory technician provided direct instruction to the researcher in all practical aspects of the saliva collection and biomarker assaying. The clinical biochemist supervised the analysis of the data set, ensured that the precision of the researcher's laboratory work was adequate and insisted that the samples were analysed in duplicate. These supports ensured that the sCort and sAA data were of highest precision, accuracy and therefore validity. Further, the validity of the high sCort concentration and sAA activity variability detected between groups.

5.6 Summary

The results of Study 2 identified the presence of a relationship between music listening and arousal consistent with the theoretical three-stage mediating model proposed. In addition, the sCort and sAA data presented novel findings in relation to how boys with LFA responded biologically to the *Sonata Pathetique* as they were exposed to a TSST-C demand condition in the form of a simulated morning school bus ride. Practically, the implications of reducing arousal for boys with LFA in other controlled but demanding environments may be of use to both them and their primary carers for service value, health outcomes and patient compliance. Further, the results of the present controlled study provide the impetus for applying Study 3 which investigated the potential for music listening to reduce arousal during real school bus rides. Specifically, it enabled the assessment of a hypothesis that reducing biological arousal may assist in the reduction of SIB for boys with LFA.

CHAPTER 6: Study 3

It is not uncommon for research designed to treat self-injurious behaviour (SIB) to use a single-case design (Banda, McAfee, & Hart, 2009; Campbell, 2003; Ford, 1999; Jennett et al., 2011; Tate & Baroff, 1966; Wachtel et al., 2009; Wachtel et al., 2011). This type of research is common in the field of psychology, is time-efficient, cost effective and can minimise risks of harm to children before larger cohort studies are conducted (Lange & Lainhart, 2009; Shadish, 2012; Yin, 2003). Unfortunately, naturalistic research designed to treat SIB exhibited by those with ASD can be difficult to administer and often goes unreported (Healey et al., 2001). However, research designed to assess children's physiological reactions to demand conditions can shed light on how they regulate arousal (Bauer et al., 2002). In addition, Ford (1999) suggested that future research may investigate the possible effects of listening to music as a way of treating SIB on transport such as buses.

Based on the above rationale, Study 3 used a single-case design comprising two conditions with repeated trials to measure the biological arousal and SIB of three school-aged boys with LFA as they took their usual morning school bus ride. Registered as a clinical trial with the Australian New Zealand Clinical Trials Registry (ANZCTR: 12611001064998) as an ethical requirement, this study tested the validity of the three separate but interconnected components of a theoretical three-stage mediating model. As seen in Figure 6.1, the theoretical model investigated the potential for biological *arousal (*measured by *sCort & sAA)* to mediate a relationship between *music listening* and *SIB* among boys with LFA exposed to a naturalistic setting provided by their usual morning school bus ride.

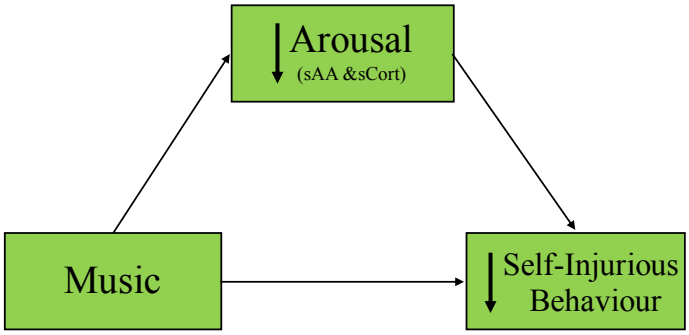


Figure 6.1 Study 3: Three-stage mediating model.

Study 3 tested numerous hypotheses within the model. Hypothesis 1 (H1) predicted that listening to calming music would reduce the frequency of SIB (ear-

blocking or pica) compared to non-exposure amongst boys with LFA who exhibit SIB. Hypothesis 2 (H2) predicted that listening to calming music would be associated with a reduction sCort concentrations compared non-music exposure. Hypothesis 3 (H3) predicted that listening to calming music would be associated with a reduction in sAA activity compared to non-music exposure. Hypothesis 4 (H4) predicted that reductions in sCort concentrations would be associated with reductions in SIB frequencies. Hypothesis 5 (H5) predicted that reductions in sAA activity would be associated with reductions in SIB frequencies. Hypothesis 6 (H6) predicted that changes in sCort concentrations would be associated with changes in sAA activity. Hypothesis 7 predicted that increases in SIB would be associated with either a symmetrical association or an asymmetrical association between sCort concentrations and sAA activity (Bauer et al., 2002). The above hypotheses were applied to the sCort concentration, sAA activity and SIB frequency data obtained from Bill, Harry and George (not their real names), three boys with LFA who exhibited SIB.

Moreover, the theoretical three-stage mediating model would be supported if music listening was associated with reduced sCort concentrations and sAA activity on the actual morning school bus ride, and these reductions were associated with reduced SIB frequencies. If this occurred, sCort concentrations, sAA activity and SIB frequencies would fluctuate in tandem across the four treatment days. Specifically, and in the convention of singe-case designs, sCort concentrations, sAA activity and SIB frequencies would decrease from Day 1: No Music to Day 2: Music, increase from Day 2: Music to Day 3: No Music, and decrease from Day 3: No Music to Day 4: Music (Kennedy, 2005).

6.1 Method

6.1.1 Participants

Three school-aged males aged 16 years, 16 years and 14 years with LFA and SIB were purposefully selected for inclusion via special education schools, the Public Transport Authority – School Bus Services (PTA-SBS) and the snowballing recruitment method employed in Study 2. The participant inclusion criteria, saliva sampling protocol and music listening condition were identical to that of Study 2 (Berk, 2007; Lopata, Volker, Putnam, Thomeer, & Nida, 2008; Tate & Baroff, 1966; Wachtel et al., 2009; Wachtel et al., 2011). In addition, the participants were required to regularly access the PTA-SBS morning school bus service and exhibit SIB often enough to be observed. Participants were excluded from the study if, in the opinion of

their primary carers, they were likely to have an aversion to music listening, the saliva collection protocol or wearing SIB personal protective equipment if previously prescribed.

Bill and George accessed PTA-SBS school bus services managed by an independent departmental contractor. Both boys travelled to school in a 21-seat commuter bus. Harry accessed morning school bus transport services from a private taxi company which operated a 12-seater bus. Harry's primary carers had come to this arrangement after numerous allegedly physical altercations with students and staff on a school bus operated by an independent education department contractor. The following apparatus were specifically sourced, designed, tested and used to record footage of Bill, Harry and George's behaviour, rate the frequencies of their SIB and collect their saliva samples on the school bus and at school across the four days of treatment.

6.1.2 Apparatus

6.1.2.1 Camera box

To record footage of participant behaviour on the school bus, a small video camera was embedded within a plastic drink bottle crate (see Figure 6.2). The crate was covered with a black blanket to make it more unobtrusive. Once operational, the crate was positioned in various locations on school buses to record participant behaviour (see Figure 6.3). Adhesive tape was used to affix the camera box to these locations to ensure that it did not move. A boom microphone was attached to the top of the camcorder to record audio of higher quality than the standard inbuilt camera microphone. Testing of the camera box revealed that it recorded footage of sufficient quality, stability and field of view to accurately assess SIB unobtrusively.

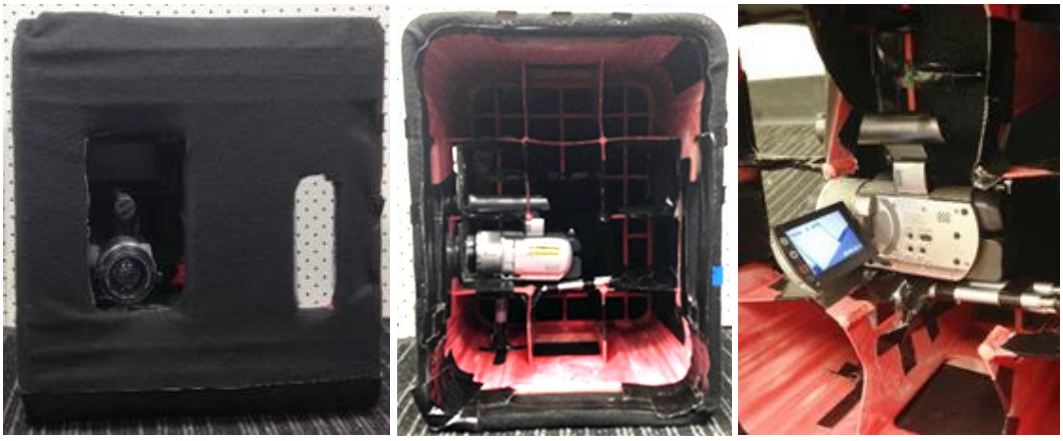


Figure 6.2 Study 3: Front, side and zoomed-in view of camera mounting.

Figure 6.3 Study 3: Camera box placement for Bill, Harry and George (left to right).

6.1.2.2 Eye glasses video recorder

To record footage of participant behaviour at school, the researcher procured a pair of *720P Eye Glasses Video Recorder* which had been manufactured by Techview (see Figure 6.4). The glasses featured a centrally located camera lens (as indicated by the orange dot in Figure 6.4), a concealed pin-hole microphone, 640 x 480 video resolution and video recordings of 1280 x 720 at 30 frames per second in high definition.

Figure 6.4 Study 3: 720P Eye Glasses Video Recorder with orange dot to indicate camera lens location.

The researcher wore the eye glass video recorder to video participant behaviour on school grounds for the 20 minutes of their school day; during the Post_5, Post_10 and Post_20 saliva collection time-points. While testing the operation of the glasses, the researcher found that the location of the camera lens was higher than his eye line. As a result, the field of view recorded was much higher than intended. In response, the researcher wore the glasses lower on his nose which recorded an appropriate field of view (see Figure 6.5).

Figure 6.5 Study 3: The researcher wearing eye glasses video recorder in the usual position and adjusted position with orange dot indication camera lens location.

6.1.2.3 Saliva collection, assays and music application

The Salimetrics Child Swabs, swab collection tubes, 1-3002 Salimetrics research Cortisol kits, 1-1902 Salimetrics Research Alpha Amylase kits, BIG Jambox manufactured by the Jawbone company and tablet computer identical to those used in Study 2 were used in Study 3. In addition, the saliva swab collection tubes were fastened to plastic ice-packs with two elastic bands to ensure that they remained cool as they were transported from the school to the laboratory for processing, as per Study 2. The sCort concentration and sAA activity analyses and intra-assay precisions reported in Study 2 also applied to the saliva samples assayed in Study 3.

6.1.2.4 Motivation assessment scale

The motivation assessment scale (MAS) by Durand and Crimmins (1988) is a seven-point Likert primary carer scale that scores responses to 16 questions and provides a possible indication as to the functional underpinnings of specific challenging behaviours such as SIB. When it was introduced, Durand and Crimmins (1988) based the reliability of the MAS on teacher and teacher assistant ratings of challenging behaviours exhibited by 50 school-aged children ranging in age from 3 to 18 years (M = 14.5 years) with ASD and other developmental disabilities. Significant inter-rater Pearson correlation coefficients ranging from .66 to .92 were reported for raw scores recorded on all 16 questions of the MAS. This indicated a high level of agreement between inter-raters, and resulted in the MAS being used frequently to identify the function of challenging behaviours exhibited by those with ID and developmental disabilities such as ASD (Joosten & Bundy, 2008).

However, re-assessments of the validity of the MAS has produced mixed results. Zarcone et al. (1991) assessed MAS ratings provided by a teacher and teacher assistant at a school, and clinical supervisor and therapy aide in an institution, to

determine the function of challenging behaviours exhibited by 55 adolescents and adults who had severe to profound intellectual disabilities including ASD. The age range of participants was not reported. A comparison of MAS responses between the inter-raters revealed only 15% agreement; well below the 80% minimum requirement for such an analysis.

Further, Sigafoos, Kerr, and Roberts (1994) assessed aggressive behaviours exhibited by 18 adolescents and adults with intellectual disabilities living in supported accommodation ranging in age from 14 to 40 years (M = 26 years). The MAS ratings provided by both a direct carer and a professional staff member achieved an overall Pearson's correlation of 34%; again well below the aforementioned minimum requirement. Despite this, as a 44.44% agreement rating was achieved by half of the sample when rating the perpetuating factor for aggressive behaviour, the researchers concluded that the MAS may be appropriate for certain participants.

Most recently, Koritsas and Iacono (2013) assessed the reliability of the MAS when they asked 52 support workers to complete the questionnaire to rate challenging behaviours of 73 adults with intellectual disabilities ranging in age from 19 to 73 years (M = 35.8 years, SD = 13 years). Cronbach's alpha coefficients conducted on the MAS total domain scores revealed internal consistencies above 0.70, suggestive of good internal consistency. Further, MAS intra-class correlation coefficients achieved over 75% agreement between inter-raters which was deemed an acceptable level of agreement. However, overall, individual item correlations, spearman rho correlations, Pearson product–moment correlation coefficient and construct validity via principle axis factoring with Varimax rotation revealed an inadequate inter-rater reliability. In sum, Joosten and Bundy (2008), reported the construct validity of the MAS to be insufficient for the assessment of challenging behaviours amongst those with multiple diagnoses as is the case for people with LFA.

6.2 Procedure

Once participants met the inclusion criteria and consent had been signed, their primary carers were interviewed by the researcher to collect information for the case presentations and to complete the Motivation Assessment Scale (MAS; Durand & Crimmins, 1988). The clinical interview was audio recorded for case presentation accuracy.

As seen in Figure 6.6, treatment days were administered on the same day of the week over four consecutive weeks. The researcher rode the school bus for the

duration of all treatments. On the two no music treatment days (days 1 and 3), when the bus was approximately 25 minutes away from arriving at school, the researcher commenced recording video footage of participant behaviour via the camera box for rating SIB frequencies. The camera lens was pointed directly and exclusively at the participant so as to not record footage of other students on the bus. Prior to signing informed consent for participation, primary carers, school principals, classroom teachers, teacher aides, bus drivers and bus aides were aware that filming on the bus and at school would occur. No children other than the participants were recorded, and footage was used for research purposes only (see Appendix R).

When the bus was approximately 14 minutes and 35 seconds away from arriving at school, the Pre saliva sample was collected. Upon arrival at school, the researcher collected the Post_0 saliva sample, activated a saliva time-point collection stop watch, ceased recording footage via the camera box and began recording the participants' behaviour over the first 20 minutes of their school day via the eye glass video recorder. Saliva samples were then collected during this first 20 minutes of the school day at Post_5, Post_10 and Post_20 time-points. The saliva samples were then conveyed to the laboratory for analysis of sCort concentrations and sAA activity. The music treatment days (days 2 and 4) observed the same process as no music treatment days, however, the boys were exposed to Beethoven's *Sonata Pathetique* for the final 14 minutes and 35 seconds of the morning school bus ride (see Figure 6.6).

The video footage recorded via the camera box and eye glass video recorder were combined to create a continuous 45-minute film. A partial interval recording (PIR) scoring matrix was created which divided the 45 minutes of footage into 270 x 10-second time increments. The researcher and 2 inter-raters then independently watched then rated the increments across the four treatment days to record the presence or absence of SIB. The inter raters were asked by the researcher to rate the SIB which had been identified by the primary carer(s) of participants only. Based on high inter-rater reliability calculations between the researcher and 2 inter-raters, and to minimise experimenter bias, the ratings of Rater A were used to determine the number of SIBs occurring in each 10-second time period for Bill, Harry and George.

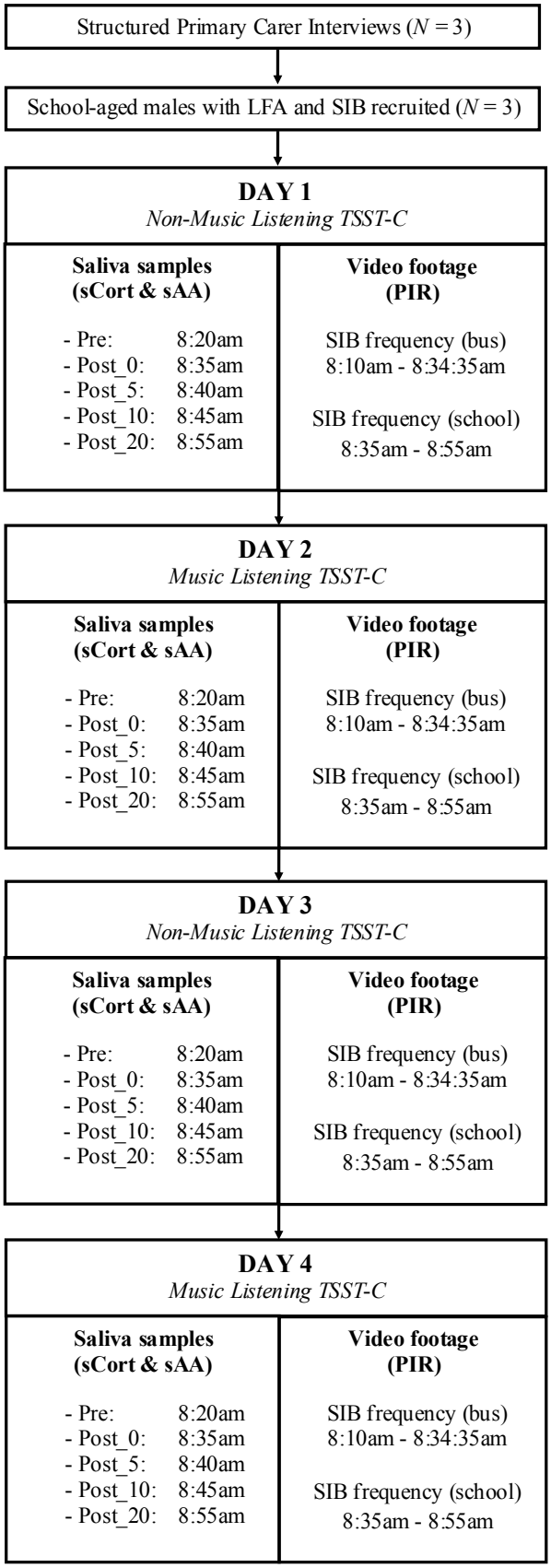


Figure 6.6 Study 3: Single-case design comprising two conditions (no music and music) with repeated trials. LFA = low functioning autism; TSST-C = trier social stress test for children; sCort = salivary cortisol; sAA = salivary alpha-amylase; PIR: partial interval recording; SIB: self-injurious behaviour.

As featured in Viau (2010), existing behavioural and pharmacological treatments undertaken by participants prior to the commencement of the present study were continued throughout the trial. This reduced the potential for extraneous variables to confound results.

The timing of the Pre-saliva sample to mark the commencement of the treatment was approximated by the school bus driver in verbal consultation with the researcher. The school buses were driven by the same drivers and attended by the same bus aides across all treatment days. The volume of the music was altered to suit the acoustics and level of sound on each treatment day as determined by the researcher in situ. The buses varied naturalistically by the number of students on any given day, hence fixing the volume of music was not possible. When the *Sonata Pathetique* was played, the bus drivers ensured that the bus radio was turned off. The music did not appear to have any adverse effects on participant behaviour or the behaviour of other students on the bus.

The researcher sat in front of the participant prior to the Pre and Post_0 saliva samples on the bus to ensure that they were not startled by his approach and subsequent saliva swabbing. Interpersonal and clinical skills were essential to the development of a working alliance with Bill, Harry and George for the saliva collection both on the bus and at school. Having previously worked as a teacher's aide at schools for students with disabilities, conducting research on the bus and within the school context was familiar to the researcher.

Over the four treatment days, the researcher rode the bus for a minimum of 45 minutes prior to collecting the Pre saliva sample and commencing video recording. This time spent on the bus promoted participant habituation to the presence and behaviour of the researcher and his materials. Further promoting this habituation, the researcher was on the bus prior to the participant boarding and wore identical clothing on each treatment day.

To ensure that the saliva was not contaminated, participants had not consumed breakfast, fluids other than water, or brushed or flossed their teeth prior to the Post_20 saliva sample on each treatment day. Consent from the primary carers indicated that this was a manageable requirement. At the conclusion of the Post_20-minute saliva sample, each participant ate their breakfast at school and was provided the opportunity to brush and floss their teeth.

Saliva samples could not be collected during the periods of music application for several reasons. Firstly, interrupting the participants during the music listening would have likely influenced their naturalistic behaviour and SIB. This may have acted as a "blocking" method (Ford, 1999, p. 302). Secondly, the presence of the researcher required to collect the sample during music listening may have influenced biological arousal.

The teacher's aides met the participant as they arrived at school by bus before accompanying them to their classroom as per school policy. Upon arrival at the classroom, the teacher's aides and teachers commenced usual classroom tasks of the day, allowing the researcher to record footage of the participant behaviour and collect saliva samples at the pre-determined time-points.

6.2.1 Ethics

Ethical approval was sought and granted by the Curtin University Human Research Ethics Committee, the Department of Education of Western Australia and the PTA-SBS of Western Australia (see Appendix R). Information letters and signed consent was sought and obtained from primary carers of participants, school principals and classroom teachers and PTA-SBS school bus contractors prior to the commencement of the study (see Appendix Q). Hard copies of the signed consent forms were stored in a locked and fireproofed cabinet at Curtin University. All electronic information such as emails, signed consent forms, sCort and sAA data, and primary carer clinical interview audio recordings were stored on a password protected computer accessed via a password protected login on the Curtin University computer server. Saliva samples were catalogued, securely stored, and de-identified as per the Study 2 protocol.

The researcher administered all treatment days and held a current Senior First Aid certificate, an unencumbered *Working with Children Check Card* and *National Police Clearance* for the duration of the study. All consenting parties were made aware by the researcher that withdrawal at any time without prejudice or penalty was supported. The researcher was prepared to cease the music and stop recording SIB attempts if participant behaviour negatively impacted on the participant or others on the school bus or at school.

6.2.2 Participant recruitment via special educational schools

The researcher accessed contact details of the schools from the Department of Education Government of Western Australia website:

http://www.det.wa.edu.au/schoolsonline/home.do. The researcher then telephoned school principals to discuss Study 3 and offered information letters for primary carers of potential participants. In instances where the school principal agreed to participate, an information letter was sent via email and a consent form was printed, signed, scanned and return emailed to the researcher (see Appendix Q). Upon receipt of the signed consent form, the researcher then arranged a time to meet the principal at the school and provide hard copies of teacher and primary carer information letters and consent forms. Principals were also provided with a recruitment Flyer upon request (see Appendix O). School principals delivered hard copies of these documents face-to-face to primary carers of potential participants at school. The primary carers were then invited to make direct contact with the researcher. Girls were excluded due to the possible variance in salivary biomarkers of arousal which can be influenced by the menstrual cycle (Bitsika et al., 2014).

6.2.3 Participant recruitment via the PTA-SBS

Based on the information provided by the researcher to gain ethical approval via the PTA-SBS of Western Australia ethics committee, the authority identified a list of potential participants. As requested by the PTA-SBS, the researcher provided hard copies of the participant information letters and consent forms. These hard copies were mailed directly to the primary carers of potential participants by the PTA-SBS on the researcher's behalf. The PTA-SBS included a cover letter which explained an opt-out recruitment option. As such, the primary carers of potential participants were availed a two-week period from the estimated date of receiving the letter and consent form to inform the PTA-SBS that they did not want the researcher to be provided with their contact details. After the opt-out period had passed, the PTA-SBS compiled a list of primary carers of potential participants and their contact details. The researcher was then invited to the state office of the PTA-SBS to access the list and contact the primary carers of potential participants under the proviso that this information was not to leave the venue. The researcher attended the PTA-SBS and contacted the primary carers of potential participants via telephone and email to inform them of the study and answered any questions in relation to inclusion and participation in Study 3.

A total of nine potential participants were identified across four special education schools and the PTA-SBS. Of these nine, five were deemed to meet the initial inclusion criteria and were accepted into the study. The researcher then visited

schools and informally observed the potential participants for the presence of SIB. As a result, three boys were deemed to be eligible for inclusion: Bill, Harry and George.

6.3 Design

A "case presentation" format was used to present the single-cases of Bill, Harry and George (Wachtel et al., 2011, p. 147). In addition, single-case design resources were reviewed to ensure that the presentation of cases and results met conventions (Cooper, Heron, & Heward, 2007; Kennedy, 2005). The information contained within each case was obtained from clinical research interviews with primary carers of participants prior to the commencement of the research. The participants were identified by their schools as being appropriate for inclusion based on their observable and identifiable SIB. Primary carers confirmed their son's LFA diagnosis by signing informed consent prior to participation. The clinical interview protocol was derived from the content of both historical and contemporary publications reporting information regarding ASD and SIB. Regarding ASD, the single-cases of Donald T. as published in *Autistic Disturbances of Affective Contact* by Kanner (1943, pp. 217-222) and Fritz V. as published in *Autistic psychopathy' in childhood* by Asperger (1944, as cited in Frith, 1991 pp. 39-50) and the DSM-5 diagnostic criteria for ASD (American Psychiatric Association, 2013) were reviewed. Regarding SIB, single-case designs published by Ford (1999) and Wachtel et al. (2011) were reviewed. See Appendix T for the final clinical interview protocol. The administration of the interview was audio recorded then reviewed by the researcher for relevant information which formed the single-cases for each participant.

In addition, the MAS by Durand and Crimmins (1988) was administered to primary carers of participants. MAS results are included in the case presentations. The specific challenging behaviour for the MAS was the SIB identified by the primary carers of Bill, Harry and George as the focus of Study 3.

The frequencies of each participant's SIBs (ear-blocking or pica) and each participant's sCort concentrations and sAA activity were periodically recorded for each of the four treatment days on the same day each week over four consecutive weeks.

6.4 Data analysis

6.4.1 Hypotheses

Table 6.1 defines hypotheses formulated for each participant of Study 3.

Table 6.1

Study 3: Analysis Stages and Hypotheses

Analysis	Hypothesis	Definition
Stage 1	H1	Listening to calming music will reduce the frequency of SIB (ear-blocking or pica) compared to non-exposure.
	H2	Listening to calming music will be associated with a reduction sCort concentrations compared non-exposure.
	H3	Listening to calming music will be associated with a reduction in sAA activity compared to non-exposure.
Stage 2	H4	Reductions in sCort concentrations will be associated with reductions in SIB frequency.
	H5	Reductions in sAA activity will be associated with reductions in SIB frequency.
	H6	Changes in sCort concentrations will be associated with changes in sAA activity.
	H7	Increases in SIB will be associated with either a symmetrical association or an asymmetrical association between sCort concentrations and sAA activity (Bauer et al., 2002).

6.4.2 Stage 1

Stage 1 of the data analysis (H1, H2, and H3) involved testing the effects of music on the frequency of SIBs (ear-blocking or pica), sCort concentrations, and sAA activity. These hypotheses were assessed via relatively simple visual techniques. If the visual analyses showed relatively weak effects in favour of the hypotheses, then they were subjected to further statistical tests. Given the high inter-rater reliabilities (see Tables 6.2, 6.4, & 6.6) between the SIB 10-second frequencies recorded by the researcher and the two independent raters (Raters A and B), the ratings of just one rater (Rater A) were used in the analyses. Before assessing the possible effects of music listening on SIB frequencies, it was essential to establish reliable SIB frequency baselines. Baseline estimates should be recorded in the same environment of music listening (on the bus), and be relatively stable across time. Relatively stable SIB baselines were observed for Bill and George, but not Harry (see Figures 6.9, 6.10, 6.14, 6.15, 6.19, & 6.20). The SIB 10-second frequency ratings of the researcher were not

used to eliminate experimenter bias. For each participant, sCort concentrations and sAA activity were standardised into z-scores to enable comparability. The mean and standard deviation (*SD*) of the participant's sCort concentrations and sAA activity was calculated across the four treatment days of Study 3 (see Figures 6.11, 6.12, 6.16, 6.17, 6.21, & 6.22).

6.4.3 Stage 2

The second stage of the analysis (H4, H5, H6, H7) was a departure from the conventional single-subject analysis as it tested possible associations between sCort concentrations and SIB frequencies (H4), sAA activity and SIB frequencies (H5), sCort concentrations and sAA activity (H6), and joint sCort concentrations/sAA frequencies and SIB frequencies (H7). For each of the three participants, these associations were tested using a series of Fisher's Exact tests. SIB frequencies, sCort concentrations, and sAA activity were converted into categorical variables using the following procedures. The sCort concentrations and sAA activity z-scores were categorised as either a positive z-score or negative z-score (the few z-scores equalling zero were excluded from the analysis). For each participant, pairs of sCort concentration and sAA activity z-scores across the four treatment days were each categorised as either both positive, both negative, sCort concentration positive/sAA activity negative, or sCort concentration negative/sAA activity positive. Lastly, SIB frequencies across the four treatment days were categorised as 0% 10-second interval occurrence per minute, between 0 and 99% 10-second interval occurrences per minute, or 100% 10-second interval occurrence per minute. The relationships among these categorical variables were evaluated with Fisher's Exact tests. The Fisher's Exact tests were preferred to chi-square tests due to the small number of observations in each cell. Because sCort concentrations, sAA activity and SIB frequencies were measured across multiple treatment days (Day 1: No Music, Day 2: Music, Day 3: No Music, and Day 4: Music) and in different conditions (on the bus with music, on the bus without music, and at school), there was expected to be sufficient variability in the measures for relationships to emerge.

6.5 Single-case presentation: Bill

The information contained in the following case presentation was obtained from the clinical research interview administered by the researcher to Bill's primary carer. As a result of Bill's status as a state ward, information about his early developmental period was not available. To retain Bill's anonymity, other than this not

being his real name, some additional information has been altered. However, the essential information has been retained for clinical relevance. Further, the content of Bill's case presentation was endorsed by his primary carer via a member checking procedure.

6.5.1 Family and developmental history

Bill was born in 1997 on the east coast of Australia. Up until 10 years of age, Bill lived with his biological mother and brothers. At this age, Bill moved to Western Australia with his mother and brothers as his biological father was estranged from the family. Not long after moving, Bill's support needs came to be in excess of what his family could provide. As a result, his mother placed him into state care and returned to the east coast. Bill's family rarely visited or contacted him. Bill met all of his childhood developmental milestones apart from a complete absence of speech.

6.5.2 Accommodation and schooling

Bill lived between two supported accommodation placement homes that provided 24 hours per day support for people with severe disabilities. Prior to 10 years of age, Bill was schooled at home by his biological mother. However, since being placed into state care, Bill had attended specialised schools for students with disabilities. At school, Bill participated in a modified school curriculum. Within this, his proficiency lay in sorting objects of various colours and shapes with the support of a full-time education assistant.

6.5.3 Physical attributes and demeanour

Sixteen-year-old Bill was 177 cm tall and weighed 75 kg at the time of this research. He had a muscular physique, pale skin complexion with severe acne scars, and short, dark brown hair. His feet often turned outwards as he swayed from side to side when he walked. He was tactile defensiveness and often adopted a rigid body posture.

Bill presented with a pleasant, timid, introverted but curious demeanour. His vision, hearing, fine and gross motor skills were typical. At school, he exhibited an extraordinary spatial awareness as he rocked back and forth while flapping his arms in crowded spaces without coming into contact with others. Bill isolated himself in social environments, preferring to rock back and forth whilst blocking his ears and producing a low frequency humming sound in the corner of a room. At his accommodation, Bill was known to be inquisitive and eager to explore without undue anxiety.

Bill's predominant facial expression was flat. When Bill appeared to be overly excited or frustrated, he lunged backwards and forwards with pursed lips producing a whistling sound whilst flapping his hands and arms vigorously. No indication of traumatic brain injury was reported or detected nor had a special interest emerged.

6.5.4 Communication

Bill did not use functional verbal language to communicate. Instead, he used non-verbal communication such as gestures and changes in facial expression to indicate pleasure or displeasure. Bill's non-verbal communication was mostly centred on having his immediate needs for food and personal care met.

Bill also used a variety of vocal tones combined with non-verbal gestures to communicate. Displeasure was often communicated by a loud and low pitched guttural sound combined with a pressured physical approach with both hands raised high above his head. Alternatively, Bill communicated pleasure by producing a soft, high pitch trill vocalisation whilst smiling and laughing. Bill responded to his name but this was often latent.

6.5.5 Nutrition

Bill preferred eating foods that produced audible crunching sounds such as rice crackers and avoided those that did not, such as cooked meat and vegetables. He also preferred foods that he could consume at speed such as sultanas and yoghurt. Despite these food preferences and aversions, Bill ate a balanced diet thanks to his carers who often disguised avoided foods in crunchy toasted sandwiches. Instrumentally, Bill consistently requested food by presenting a dinner plate to primary carers. Bill preferred carbonated drinks and often refused to consume other beverages.

6.5.6 Music

At his supported accommodation, when exposed to fast tempo music, Bill danced by lunging backwards and forwards whilst flapping his hands and arms with elbows raised. Continuous dancing in this manner resulted in Bill becoming over aroused and physically aggressive toward others. To limit this behaviour, Bill was discouraged from listening to music. Based on this response to music, Bill's primary carer was unsure if he would tolerate listening to the *Sonata Pathetique* during his morning school bus ride.

6.5.7 Behaviour

Bill's primary carer believed that he would not accept the saliva collection swab due to an established oral tactile defensiveness. For example, Bill frequently

refused support to shower, bathe and brush his teeth. When approached by staff to complete these tasks, Bill retreated to the corner of the room and screamed loudly. When displeased or frustrated, Bill became physically aggressive toward carers. Further, he often woke numerous times each night, to disrobe and stand facing the corner of his bedroom.

Bill required staff support to complete simple daily tasks such as washing, dressing, socialising, eating, and transitioning between tasks. He ate food with his fingers as opposed to utensils, had not yet achieved toileting proficiency. In sum, Bill was globally delayed in most aspects.

6.5.8 Behavioural interventions and medications

Bill had not undergone behavioural treatments. However, he was taking a morning and evening dose of risperidone to reduce the emergence of aggressive behaviour.

6.5.9 DSM-5 ASD features

Bill's primary carer confirmed that he had official diagnoses of ASD and ID which met the criteria for LFA and resulted in significant social, academic and daily impairments. His carer described him as introverted but socially curious with a reliance on non-verbal communication in the absence of a functional verbal language. Bill exhibited various stereotypical body movements, the most frequently occurring being ear-blocking SIB.

Regarding the first domain of the DSM-5 diagnostic criteria for ASD, Bill presented with severe deficits in social communication, interaction, reciprocity, approach, interests and sharing of interests, conversation, initiation, response, developing and maintaining relationships, adjusting behaviour to different social environments and play. He engaged socially to have his immediate needs met. In the classroom, Bill responded well to Auslan keyword sign language prompting for "work", "toilet", "more", "one more", "please", "good" and "thank you". He appeared to be aware of others, however he did not possess the social skills to engage.

Regarding the second domain of the DSM-5 diagnostic criteria for ASD, Bill had an over-reliance on routine, sensory seeking such as carrying small objects in the palm of his hand as well as laying under heavy couch cushions and wrapping himself tightly in blankets for deep pressure sensory input. He enjoyed the vestibular sensation of rocking and lunging. He also exhibited repetitive motor movements and patterns of

behaviour such as hand posturing, body rocking, finger play, self-stimulation and SIB in the form of ear-blocking.

6.5.10 SIB: Ear-blocking

Bill's ear-blocking SIB involved him using the distal phalanx of his index and middle fingers to put heavy pressure on the Tragus whilst the distal phalanx of the thumb also provided pressure from underneath the lobule. This action involved both ears and resulted in the External Acoustic Meatus and Tympanic Membrane being completely blocked (see Figure 6.7).

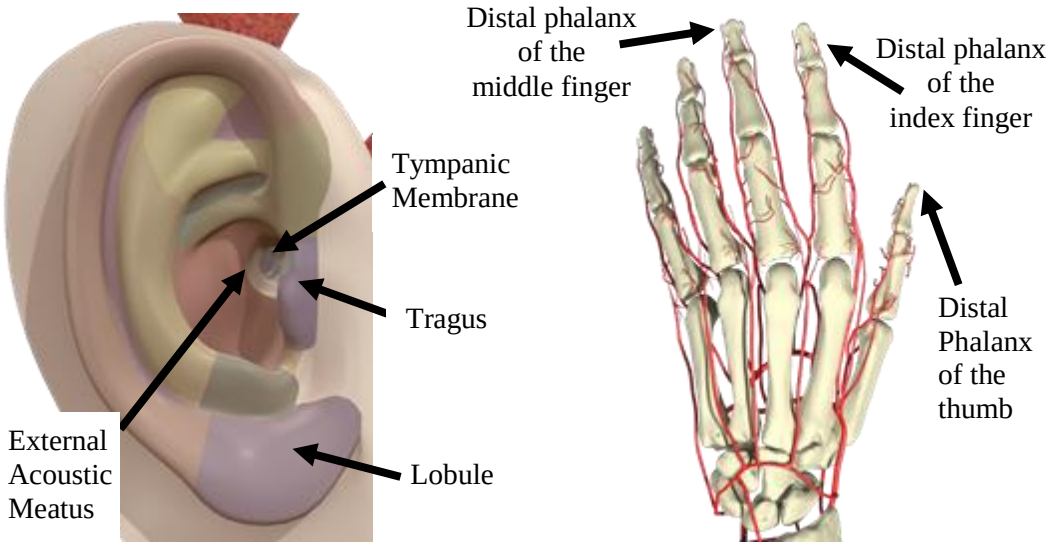


Figure 6.7 Study 3: External anatomy of the ear and the skeletal structure of the phalanges of the human hand (Primal Pictures, 2006b, 2006c).

Bill's ear-blocking SIB was pervasive, self-injurious, predictable, and persistent across environments and without a clear precipitant. However, he applied more pressure to his ears when displeased. Bill did not consistently block his ears when completing favoured classroom tasks, however, this was inconsistent. As a result of his ear-blocking SIB, the Tragus and beneath the lobules on both ears were red in colour and had numerous unhealed scratches created by blocking with long fingernails. Bill's carers were often unable to trim his fingernails due to refusal resulting from tactile defensiveness. Bill's ear-blocking SIB injuries caused him physical pain, infections and had the potential to cause long-term damage to the structures of his inner and outer ears. Despite this, few treatments had been attempted. Attempts to apply personal protective equipment were refused by Bill due to his tactile defensiveness.

6.5.11 Results

6.5.11.1 SIB function

A MAS was administered to Bill's primary carer to determine if the function of his SIB on the school bus and at school was motivated by attention, tangible objects, avoidance or sensory domains (see Appendix V; Joosten & Bundy, 2010). An analysis of his MAS data ranked his ear-blocking SIB as being motivated by a sensory function (see Figure 6.8).

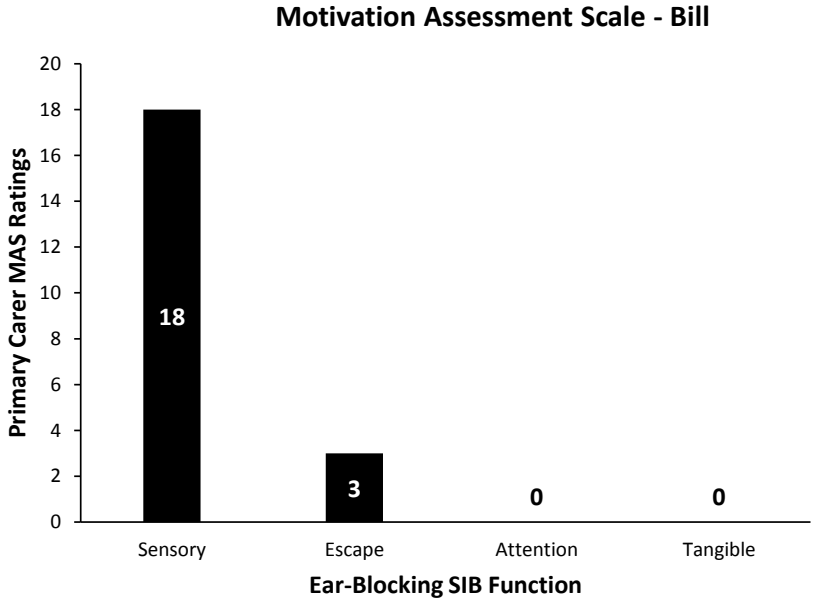


Figure 6.8 Study 3: Bill's MAS results.

6.5.11.2 Inter-rater agreement

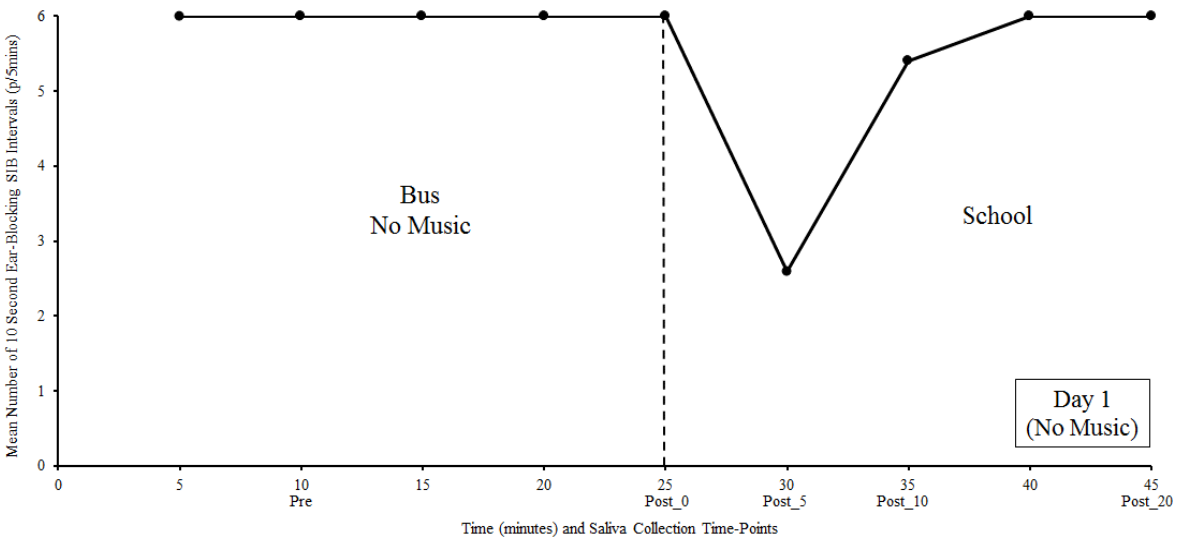
Inter-rater agreement for footage recorded on the school bus and on school grounds was assessed by the researcher and two independent and qualified inter-raters: Inter-rater A and Inter-rater B (see Appendix X). The number of 10-second intervals per minute within which Bill exhibited ear-blocking SIB were assessed across the four treatment days. As a result of the high inter-rater reliability, the interval ratings recorded by Inter-rater A were used in the analysis (See Table 6.2).

Table 6.2
Study 3: Bill's Inter-rater Agreement

Treatment Days	Inter-rater Agreement (the proportion of 10-second SIB interval ratings for which all inter-raters agreed)
Day 1 (No Music)	94.2% (130/138)
Day 2 (Music)	95.9% (190/198)
Day 3 (No Music)	100% (207/207)
Day 4 (Music)	97.6% (202/207)
Mean:	96.9%

6.5.11.3 Music listening and SIB

H1 (listening to calming music will reduce the frequency of SIB compared to non-exposure) was tested via visual inspections of Figures 6.9 and 6.10. The plots clearly show that the frequency of Bill's SIB remained at a relatively high level during all the bus rides, regardless of exposure to music. For Bill, therefore, H1 is not supported. Upon arriving at school, the frequency of Bill's SIB decreased on Day 1 (no music on bus) and Day 2 (music on bus) before returning to baseline. On Day 3 (no music on bus) and Day 4 (music on bus), Bill's SIB stopped upon arriving at school and tended to stay that way for the duration of the observation period. Because the reductions in SIB frequencies occurred regardless of whether he had been exposed to the music on the bus or not, it can be concluded that the music had neither an immediate nor delayed effect for Bill. As such, at school, no consistent SIB trends emerged on music or no music treatment days.



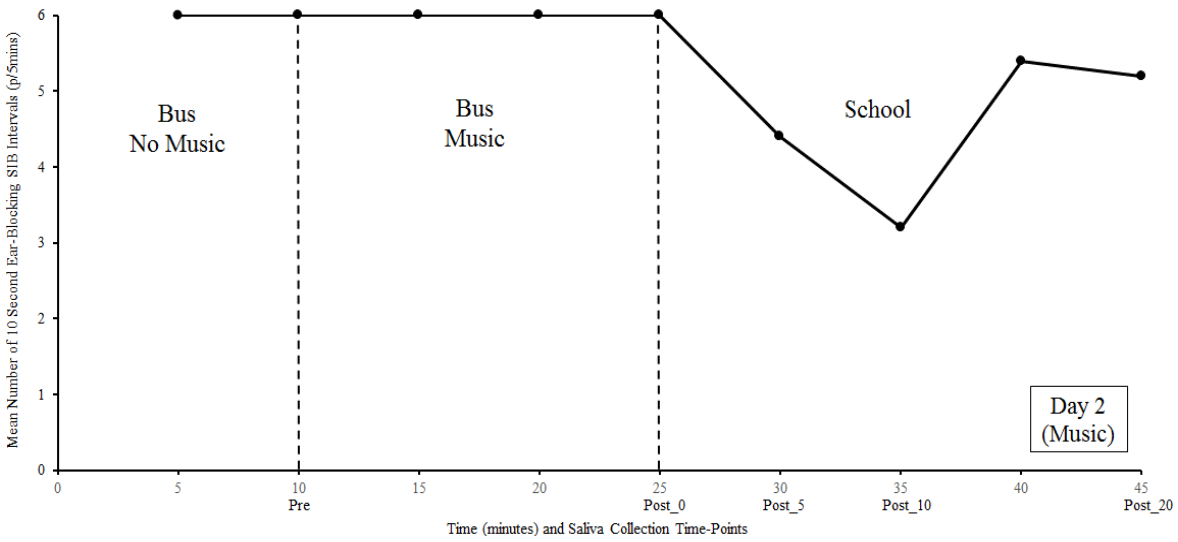


Figure 6.9 Bill's mean number of ear-blocking SIB intervals (*y axis*) with saliva collection time-points (*x axis*) for Day 1 (No Music) and Day 2 (Music) of Study 3.

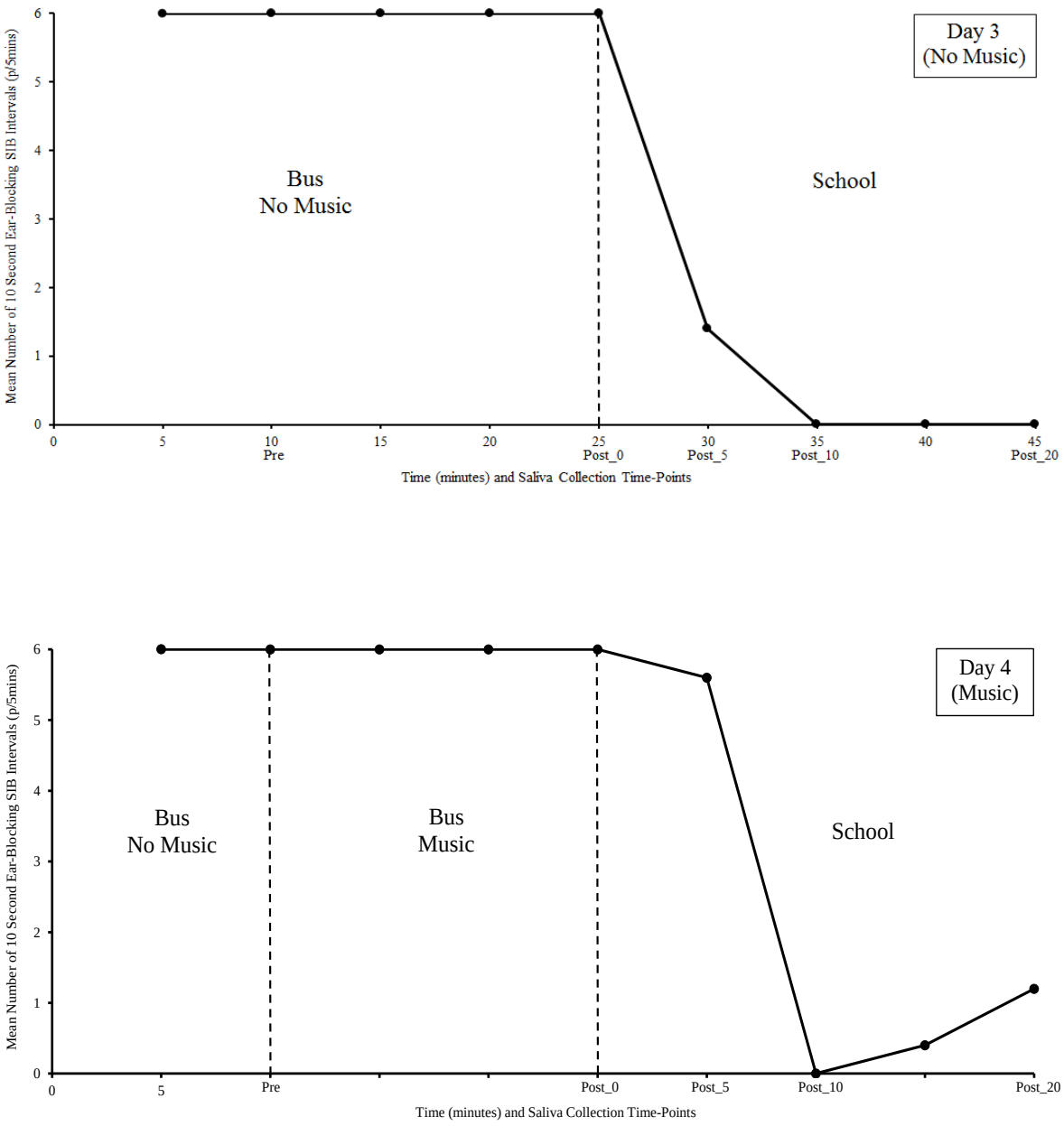


Figure 6.10 Bill's mean number of ear-blocking SIB intervals (*y axis*) with saliva collection time-points (*x axis*) for Day 3 (No Music) and Day 4 (Music) of Study 3.

6.5.11.4 Music listening, sCort and sAA

H2 (listening to calming music will be associated with a reduction sCort concentrations compared to non-exposure) and H3 (listening to calming music will be associated with a reduction sAA activity compared to non-exposure) could not be subjected to a rigorous test because Bill's saliva was not collected during periods of music listening. Collecting the saliva samples at this time was likely to have interfered with Bill's natural ear-blocking behaviour and potentially influence his sCort concentration and sAA activity readings. Therefore, the analysis of Bill's sCort concentrations, sAA activity and joint sCort/sAA concentration/activity on the bus were restricted to the standardised sCort and sAA z-score measures of saliva analysed at the Pre and Post_0 saliva collection time-points. As such, the following results should be viewed as suggestive only. The standardised scores are reported in Table 6.3.

Table 6.3

Bill's Standardised sAA Activity and sCort Concentrations Across the Five Saliva Collection Time-points for the Four Days of the Study

Treatment Day	Condition	Time-Point	sAA (U/mL)	sCort (µg/dL)
Day 1 (No Music)	Bus No Music	Pre	-0.66	0.84
	Bus No Music	Post_0	0.80	-0.02
	School	Post_5	-0.31	-1.07
	School	Post_10	-1.18	-0.67
	School	Post_20	0.44	-0.97
Day 2 (Music)	Bus No Music	Pre	0.17	2.29
	Bus Music	Post_0	-1.58	-0.04
	School	Post_5	-0.87	0.37
	School	Post_10	1.83	-0.77
	School	Post_20	-0.24	-1.70
Day 3 (No Music)	Bus No Music	Pre	0.99	2.34
	Bus No Music	Post_0	1.41	0.60
	School	Post_5	1.66	0.54
	School	Post_10	-0.19	0.38
	School	Post_20	-0.72	0.08
Day 4 (Music)	Bus No Music	Pre	-0.81	0.73
	Bus Music	Post_0	0.10	-0.25
	School	Post_5	0.18	-0.43
	School	Post_10	0.51	-0.80

Visual inspections of Figures 6.11 and 6.12 show a pre-post decline in sCort concentrations in the non-music conditions of Day 1 and Day 3 and the music conditions of Day 2 and Day 4. Moreover, the rate of decline is comparable across the conditions. H2 is therefore not supported by Bill's data.

Visual inspection of Figures 6.11 and 6.12 show a pre-post decline in sAA activity for one of the two music conditions (Figure 6.11); however, this effect was not replicated in the other music condition (Figure 6.12), which showed a pre-post increase in sAA activity. The two no music conditions of Day 1 and Day 3 both showed pre-post increases in sAA activity. H3 is therefore not supported by Bill's data. At school, there was a consistent decrease in sCort concentrations but no consistent pattern in sAA activity across treatment days.

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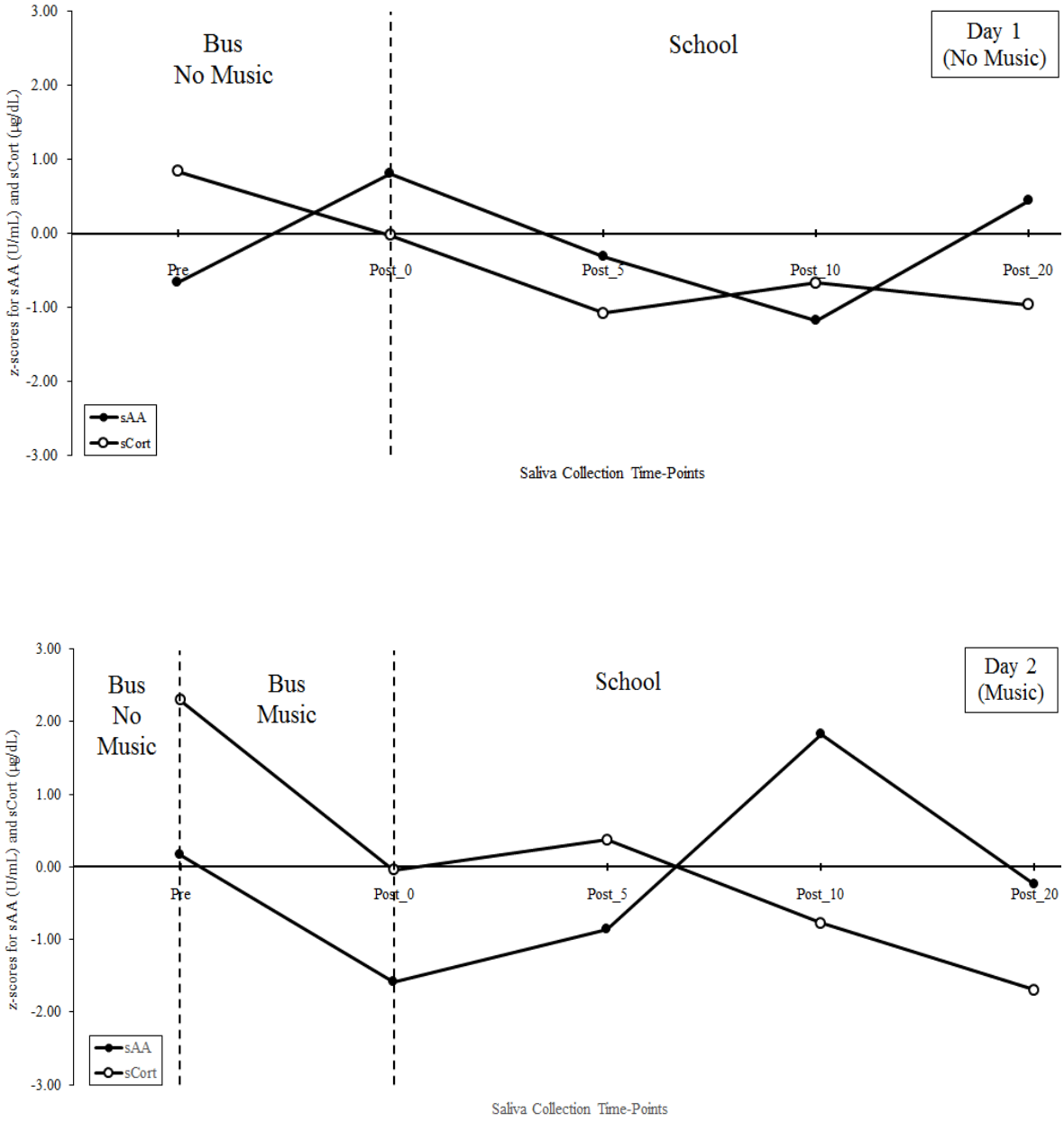
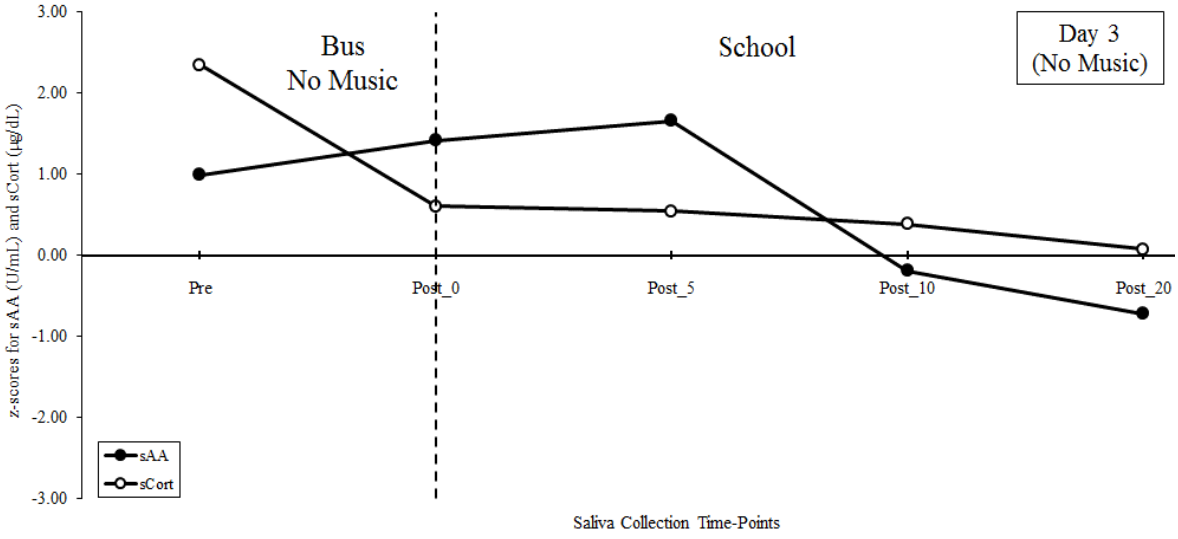


Figure 6.11 Bill's standardised sAA activity (U/mL) and sCort concentrations (µg/dL) (*y axis*) with saliva collection time-points (*x axis*) for Day 1 (No Music) and Day 2 (Music) of Study 3.



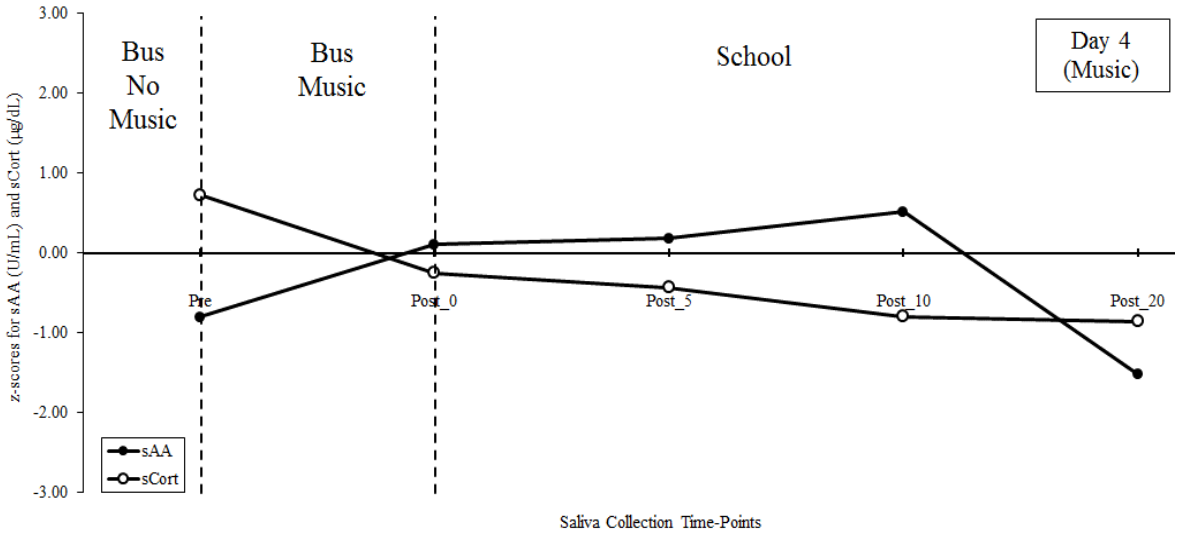


Figure 6.12 Bill's standardised sAA activity (U/mL) and sCort concentrations (µg/dL) (*y axis*) with saliva collection time-points (*x axis*) Day 3 (No Music) and Day 4 (Music) of Study 3.

6.5.11.5 SIB, sCort and sAA

H4 (reductions in sCort concentrations will be associated with reductions in SIB frequencies), H5 (reductions in sAA activity will be associated with reductions in SIB frequencies), H6 (changes in sCort concentrations will be associated with changes in sAA activity) and H7 (increases in SIB will be associated with either a symmetrical association or an asymmetrical association between sCort concentrations and sAA activity) were tested with the Fisher's Exact test according to the procedure outlined in Section 6.4.2.

In relation to H4, there was no association between sCort levels (above the mean level, below the mean level) and SIB frequencies (0%, between 0 and 100%, 100%) across the four treatment days of the study (p = .358). The 2 x 3 cell counts and frequencies are reported in Appendix U. In relation to H5, there was no relationship between sAA levels and SIB frequency across the four treatment days of the study (p = .807). The 2 x 3 cell counts and frequencies are reported in Appendix U. In relation to H6, there was no association between sCort and sAA levels across the four treatment days of the study (p = 1.000). The 2 x 2 cell counts and frequencies are reported in Appendix U. Finally, in relation to H7, there was no relationship between joint sCort/sAA levels and SIB frequency across the four days of the study (p = .792). The 3 x 3 cell counts and frequencies are reported in Appendix U. Hypotheses 4 to 7 were therefore not supported by Bill's data.

6.5.12 Discussion

Bill exhibited constant and predictable ear-blocking SIB which resulted in tissue damage to the internal and external structures of both ears. He participated in the 4 treatment days of the single-case design comprising 2 conditions with repeated trials on the same school morning each week over 4 consecutive weeks. Bill's eligibility for inclusion into Study 3 was validated by consistently high and stable baseline ear-blocking SIB frequencies. This enabled an assessment of H1 through H7. Bill's data did not support any of the hypotheses.

Ear-blocking SIB was chosen as a target behaviour for the MAS. Bill's MAS ratings indicated that his ear-blocking SIB was motivated by a sensory factor. Unfortunately, the MAS results do not specify the particular sensory factor, although, based on anecdotal reports from his primary carer, Bill's ear-blocking SIB appears to be the result of an auditory sensory aversion. As such, it is possible that Bill's ear-blocking SIB may be a method of him averting or regulating unwanted auditory

stimulus. This may explain the finding that expose to music did not reduce the frequency of his SIB. It is also possible that Bill's ear-blocking may have been his method of enhancing the sensory input of preferred auditory stimuli. An example of this may be his preference for foods that produce an audible crunch.

Bill's sCort concentrations were not associated with his sAA activity at any of the saliva collection time-points. This result was particularly interesting as sCort concentrations have been reported to be indicative of HPA axis activity and sAA activity of ANS activity in that they share the same function, namely, to stabilise the human body in response to stress and return it to a state of homeostasis (Bauer et al., 2002; Chrousos & Gold, 1992; Rudolph et al., 2010). However, these reports were based on studies including people without LFA. More specific to LFA, Corbett et al. (2012) reported significant sCort concentration variance amongst children with ASD and concluded that this was the result of heterogeneity. Furthermore, Kidd et al. (2012) asserted that research reporting sAA activity in people with ASD did not exist. As a result, sAA has not become empirically established as an absolute marker of ANS activity among children with ASD (Quas, 2011). Despite the lack of association between sCort concentrations and sAA activity in this study, the result may add to the body of knowledge regarding the HPA axis, ANS and their representative biomarkers of arousal amongst children with LFA.

Bill's sCort concentrations and sAA activity were not found to be associated with his ear-blocking SIB frequencies. This result does not corroborate with Jennett et al. (2011) who reported that an increased rate of SIB was linked to elevated levels of arousal for a 17-year-old female with LFA. However, the Jennett et al. (2011) study reported physiological arousal in the form of heart rate data and investigated head hitting, hair pulling and hand biting as SIB types. This differed from the present study in that sCort and sAA were reported as biological markers of arousal and ear-blocking was the SIB type. In addition, the participant in the study published by Jennett et al. (2011) was female and Bill was a male. More specifically, the gender of the participant in the study published by Jennett et al. (2011) may have affected the results as previously mentioned that saliva samples collected and analysed from females can be influenced by the menstrual cycle (Bitsika et al., 2014).

Bill's data did not support the additive or interactive models published by Bauer et al. (2002), based on the assumptions that his ear-blocking SIB was a challenging behaviour in that it causes scratches, infections, tissue damage to the ear

and can be a method of auditory sensory aversion. sCort and sAA did not present as consistent indicators of an arousal response to the stress of a morning school bus ride for Bill. However, as previously discussed Bill's ear-blocking SIB may have been contingent on the avoidance or regulation of auditory sensory stimulus.

6.6 Single-case presentation: Harry

The information contained in the following case presentation was gathered from the clinical research interview administered by the researcher to Harry's biological mother and father. To retain Harry's anonymity, his name has been altered. The content of Harry's case was endorsed by his primary carer via a member checking procedure.

6.6.1 Family and developmental history

Sixteen year-old Harry weighed 4.1 kilograms at birth. He lived with his mother, father, sister who had high functioning autism and brother who had Down syndrome. Equipped with a bachelor's degree in primary school teaching, Harry's mother was a primary carer for her children. Having achieved a master's degree in management, Harry's father was employed in risk management for a state government department as well as caring for his children.

Harry's prenatal period was typical. He was born via natural delivery two days past his due date and spent one week in a humidicrib to recover from low oxygen levels after birth. Harry was breast fed until 12 months of age. He was a placid baby who slept well, cried rarely, and was easily soothed. As a toddler, he achieved social, fine and gross motor skill developmental milestones within typical limits. Nurses did not raise concerns at regular child health checks.

However, at 2 years of age, Harry was overly quiet, unperturbed when separated from his parents and preferred his own company. Despite these tendencies, at 3 years of age, Harry continued to meet typical developmental milestones including age appropriate speech acquisition. Further, he showed evidence of imitating social behaviours and using social communication strategies effectively. For example, he often requested "to the park?" within context.

However, just 3 months after his third birthday, Harry's development regressed remarkably. His language regressed from words to nonsense words then to mutism. Socially, he withdrew himself completely from familiar and unfamiliar children and adults. For example, Harry refused to attend family outings in favour of reading books at home. With the origin of his regression unknown, Harry's parents sought a

comprehensive medical assessment. Subsequently, Harry was diagnosed with ASD. Harry's extended family disagreed with this diagnosis and subsequently excluded him and his immediate family from activities. This lead to Harry having fewer social developmental opportunities. Aware of this, Harry's parents enrolled him in a social skills early intervention program with limited success.

Harry achieved continence not long after he turned 4 years of age. However, this was only after an intensive period of occupational therapy intervention. However, he has not ever used the toilet outside of the family home. This has resulted in several major incidents of incontinence in public areas.

6.6.2 Accommodation and schooling

Harry attended a special education school from 5 to 6.5 years of age after one year spent at an ASD specific early intervention service focussing on speech therapy. At 6.5 years of age, as a result of school refusal, Harry was transitioned to a mainstream school. This was mildly successful for a short period of time before Harry and his family relocated due to his father's work. At 7 years of age, Harry was sent to a special education centre within a mainstream school where he remained until he was aged 10 years. At this age, his family relocated again due to his father's work. Harry was re-enrolled into a special education centre within a mainstream school. However, at 12 years of age, school refusal again resulted in him being transitioned to a special education school.

Academically, Harry's strengths were in computing and handwriting and difficulties were in mathematics and communication. Attempts to formally assess his academic and cognitive abilities were unsuccessful. His responses to verbal task requests were often delayed or absent. Harry's parents accessed government funded respite and community access programs.

6.6.3 Physical attributes and demeanour

Sixteen year-old Harry was 175 cm tall and weighed 60 kg at the time of the interview. He was of slim build with a pale complexion and black, curly hair. His visual acuity was unknown due to assessment refusal. However, his hearing ability was extraordinary. Harry's upper back was hunched due to his often bent-over posture when ear-blocking. Harry's gross and fine motor skills developed typically. He was tactile avoidant and acutely spatially aware. His walking gait appeared rushed due to him taking short and fast steps.

Harry's demeanour was rigid, solemn, intimidating, avoidant and defensive. However, he was reported to be very engaging when requiring assistance. He had not been affected by brain injury or infection.

6.6.4 Communication

After his language regressed at 3 years and 3 months of age, Harry slowly began to regain some words. Harry began to use a restricted range of keywords and short phrases such as "I cook … butter chicken", "next week … I go to … Garry's house … play piano?" Despite having the use of keywords, Harry often chose to communicate using pen and paper. When Harry did speak, it was often in first person. Harry's verbal communication was often only understood by those who knew him best. His facial expressions were limited he was often expressionless. Harry's verbal communication difficulties often resulted in frustration, anger and physical aggression toward others. Harry often reverted to physically leading people by the hand to desired items/activities. At school, Harry used a daily visual schedule to complete classroom activities.

6.6.5 Nutrition

Prior to 12 years of age, Harry ate a balanced diet. At school, he chose fruit as a positive reinforcer for the completion of work. However, after 12 years of age, Harry developed a preference for high sugar foods and drinks.

6.6.6 Music

Before the age of 12 months, Harry had a special interest in children's musical television programs. By the age of 3 years, he had learned to play single notes on a piano keyboard. At this age, Harry requested his mother play *Heart and Soul* by Hoagy Carmichael on the piano by humming the tune. At 4 years of age, and based on the promoted developmental benefits alleged by the *Mozart Effect*, Harry's mother exposed him to recordings of Wolfgang Amadeus Mozart and *Four Seasons* by Antonio Vivaldi. At 12 years of age, Harry began to listen to contemporary, heavy metal and rhythm and blues musical genres. Harry often requested radio stations featuring classical music when travelling in the family car or on the school bus. Hence, his mother predicted that he would not be averse to being exposed to the *Sonata Pathetique* during his morning school bus ride.

6.6.7 Behaviour

With close supervision, Harry was able to wash and dress himself, complete household chores and eat food that had been pre-prepared. However, the completion

of these tasks was dependent upon tangible positive reinforcers such as sugary foods. Harry had a strong aversion to barking dogs. In the presence of a dog, Harry immediately blocked both of his ears with excessive pressure as he absconded at speed. Harry was hypervigilant and unable to tolerate being in the presence of any breed of dog.

6.6.8 Behavioural interventions and medications

Despite being prescribed medicine throughout his life, Harry consistently refused. Efforts to disguise oral medications in preferred foods or drinks were also refused. Harry accessed speech therapy and physiotherapy for many years.

6.6.9 DSM-5 ASD features

Harry was tactile defensive, avoidant of loud auditory sensations, socially disconnected, and reliant on routine. He egocentrically lived in his own world where others functioned almost exclusively to meet his immediate needs. Further, he became frustrated, angry then aggressive toward others when unable to communicate effectively. Harry constantly engaged in ear-blocking SIB.

Regarding the first domain of the DSM-5 diagnostic criteria for ASD, Harry presented with severe deficits in social communication, gesture, interaction, reciprocity, approach, conversation, initiation, response and direct eye-to-eye contact. Further, he had severe difficulties detecting the emotions of others and when attempting to develop, understand and maintain relationships. He was unable to adjust his social behaviour to meet the demands of different environments or show a genuine social interest in peers. His facial expressions were limited to flat and angry.

Regarding the second domain of the DSM-5 diagnostic criteria for ASD, Harry exhibited restricted, repetitive, pervasive and predictable patterns of behaviour such as ear-blocking, lining up objects and adjusting his school bag on the hook outside of the classroom repetitively. His speech was repetitious and echolalic. For example, he immediately parroted the researcher's saliva sampling verbal prompt of "chew chew chew". He also used idiosyncratic phrases. For example, at school he would repetitively exclaim "the music flying up in the air". He displayed motor stereotypies and fixated on parts of objects as opposed to the whole. For example, he flicked and bent the dry portion of the saliva swab with his fingers before bending it to make a 90-degree angle for close visual inspection.

Harry had an insistence on sameness and rigid routines. For example, Harry became highly agitated and aggressive when he learned that a preferred carer was

resigning after 3 years. In addition, Harry constantly required social stories to forecast daily changes in routine. Harry's speech production was monotonal. For example, he uttered "Good Morning" using a low and monotonal voice across the four consecutive research mornings. Further, he often spoke using stereotypic phrases in a sing-song voice. For example, when he noticed a favoured class teacher, he exclaimed "all sings in the dirt" using a sing-song voice. Harry was avoidant of tactile and loud auditory stimuli. Examples of these included avoiding all physical contact and barking dogs, vacuum cleaners, automated hand dryers and motor driven garden machinery.

Harry's ASD features severely impacted on his residential, educational and social functioning. As a result, he required support both in the family home and school classroom. In addition, he had severe anxiety and a suspected intellectual disability. Overall Harry was a physically tall, non-complaint, avoidant and combative teenager with limited social intent or the ability to take other's perspectives.

6.6.10 SIB: Ear-blocking

The most pervasive self-injurious behaviour exhibited by Harry was ear-blocking. He used the Distal Phalanx and index finger to block the External Acoustic Meatus and Tympanic Membrane by applying significant pressure to the Targus (see Figure 6.8). Harry's ear-blocking SIB occurred at high frequency and intensity. It resulted in skin redness, skin depressions, abrasions, and infections. At 4 years of age, Harry was treated for otitis media with effusion, otherwise known as "glue ear", by grommet implants. His ear-blocking SIB emerged not long after this period.

Harry blocked his ears to avoid loud sounds such as dogs barking, loud music and power tools. Further, Harry applied additional pressure to his Targus when avoiding an unwanted task or task request. Harry's ear-blocking SIB was constant and reported to not be contingent on a single consistent factor by his parents.

Momentary departures from ear-blocking SIB were apparent when Harry was required to use one or both hands to complete a favoured task. In addition, Harry was still able to hear when ear-blocking. For example, he responded to favoured auditory stimuli when engaged in this behaviour.

Previously, it had been proposed that Harry place his hands into his trouser pockets to avert ear-blocking. Unfortunately, this intervention was unsuccessful. Further, and due to his tactile defensiveness, Harry refused to wear protective ear muffs to limit the self-injury associated with his ear-blocking. Harry had been mocked in

public about his ear-blocking. Further, his siblings were teased at school and parents were often socially excluded as a result.

6.6.11 Results

6.6.11.1 SIB function

The MAS was administered to Harry's primary carers to determine if his ear-blocking SIB on the school bus and at school was motivated by attention, tangible objects, avoidance or sensory domains (see Appendix X; Joosten & Bundy, 2010). An analysis of his MAS data ranked his ear-blocking as being highly motivated by sensory and tangible factors (see Figure 6.13).

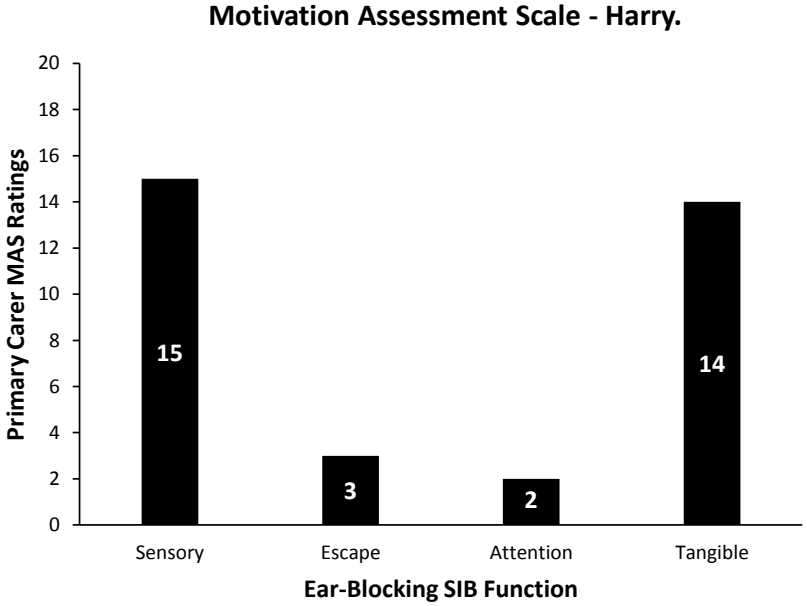


Figure 6.13 Study 3: Harry's MAS results.

6.6.11.2 Inter-rater agreement

Inter-rater agreement for footage recorded on the school bus and at school was assessed by the researcher and two independent and qualified raters as per Bills analysis (Rater A and Rater B; see Appendix X). The number of 10-second intervals within which Harry exhibited ear-blocking SIB per minute were assessed across each of the four treatment days. As seen in Table 6.4, inter-rater reliability was high across the three raters. Therefore, the SIB ratings recorded by just one rater (Rater A) were used in the analysis.

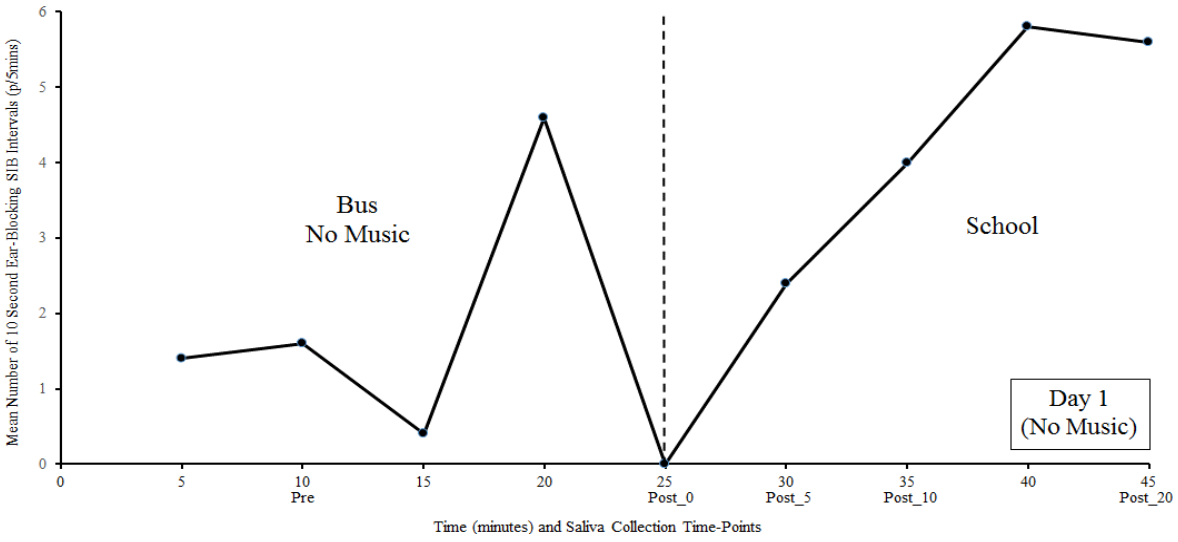
Table 6.4

Study 3: Harry's Inter-rater Agreement

Treatment Days	Inter-rater Agreement (the proportion of 10-second SIB interval ratings for which all inter-raters agreed)
Day 1 (No Music)	94.7% (196/207)
Day 2 (Music)	90.8% (188/207)
Day 3 (No Music)	96.6% (200/207)
Day 4 (Music)	96.6% (200/207)
Mean:	94.7%

6.6.11.3 Music listening and SIB

H1 (listening to calming music will reduce the frequency of SIB compared to non-exposure) was tested via visual inspections of Figures 6.14 and 6.15. Contrary to H1, Day 2 (Music) and Day 4 (Music) show increases in the frequency of Harry's SIB following the onset of the music. It would be premature to attribute these increases to music listening as similar increases in the frequency of Harry's SIB occurred on the days when there was no music on the bus. Moreover, the increases in SIB occurred at similar stages of the bus journey on music days (Days 2 and 4) and non-music days (Days 1 and 3). H1 was therefore not supported for Harry. Regardless of whether he had been exposed to music on the bus, the only common trend in the SIB trajectories during the observational periods at school was a peak during the final observational period.



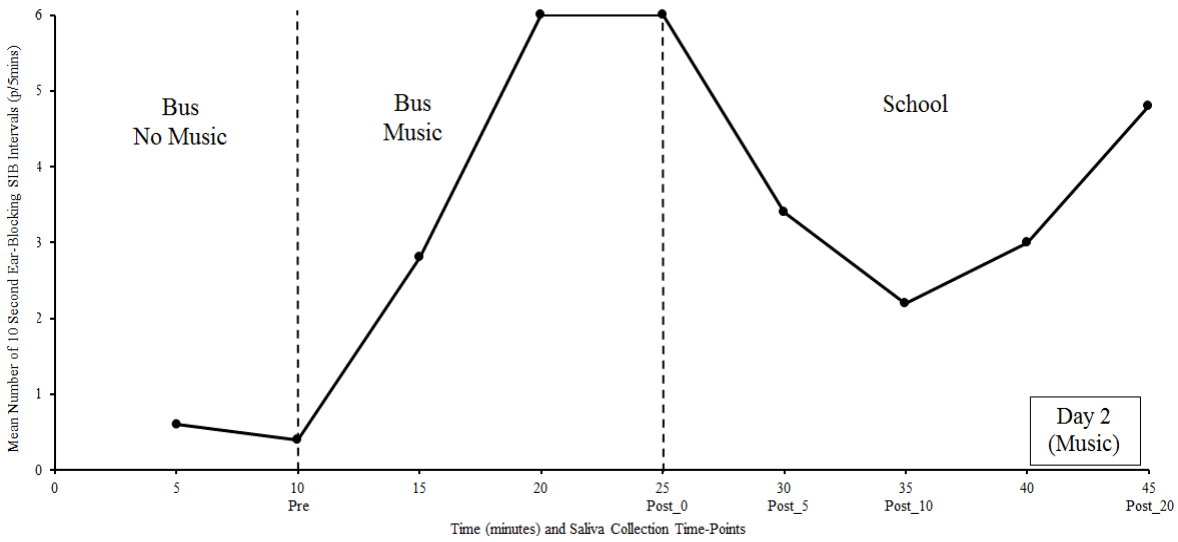
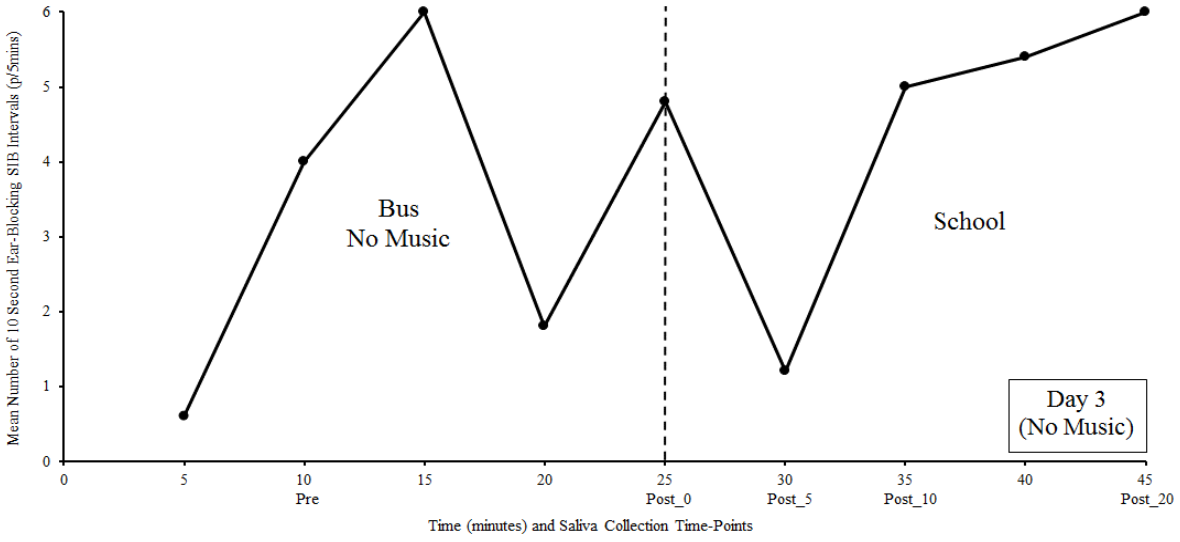


Figure 6.14 Harry's mean ear-blocking SIB intervals (*y axis*) with saliva collection time-points (*x axis*) for Day 1 (No Music) and Day 2 (Music) of Study 3.



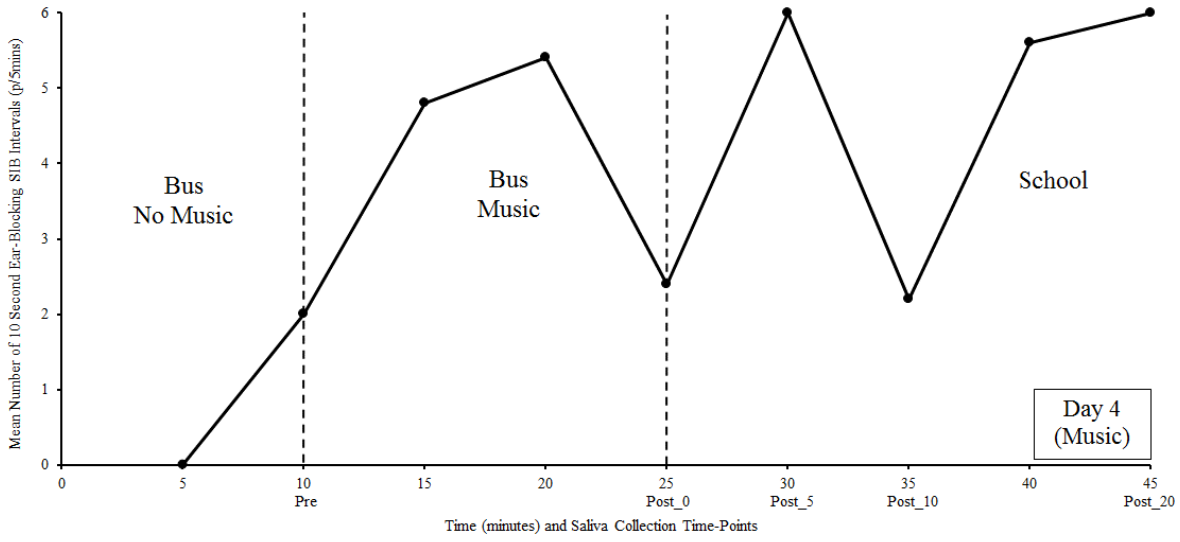


Figure 6.15 Harry's mean ear-blocking SIB intervals (*y axis*) with saliva collection time-points (*x axis*) for Day 3 (No Music) and Day 4 (Music) of Study 3.

6.6.11.4 Music listening, sCort and sAA

When testing H2 (listening to calming music will be associated with a reduction sCort concentrations compared to non-exposure) and H3 (listening to calming music will be associated with a reduction sAA activity compared to non-exposure), like Bill, the analysis of Harry's sCort concentrations, sAA activity and joint sCort/sAA concentration/activity were restricted to the standardised sCort and sAA measures taken at the Pre and Post_0 saliva collection time-points. As such, the following results should be viewed as suggestive only. The standardised scores are reported in Table 6.5.

Table 6.5

Harry's Standardised sAA Activity and sCort Concentrations Across the Five Saliva Collection Time-points for the Four Days of the Study

Treatment Day	Condition	Time-Point	sAA (U/mL)	sCort (µg/dL)
Day 1 (No Music)	Bus No Music	Pre	1.32	1.59
	Bus No Music	Post_0	0.99	0.98
	School	Post_5	1.93	Not available
	School	Post_10	0.96	-0.49
	School	Post_20	Not available	Not available
Day 2 (Music)	Bus No Music	Pre	-0.25	0.91
	Bus Music	Post_0	0.12	0.02
	School	Post_5	0.31	-0.36
	School	Post_10	0.09	-0.04
	School	Post_20	-0.57	-0.35
Day 3 (No Music)	Bus No Music	Pre	-0.77	0.65
	Bus No Music	Post_0	-0.85	0.83
	School	Post_5	-0.68	0.63
	School	Post_10	1.23	-0.32
	School	Post_20	0.44	1.03
Day 4 (Music)	Bus No Music	Pre	-0.39	0.06
	Bus Music	Post_0	-0.08	0.49
	School	Post_5	-0.41	-0.20
	School	Post_10	-0.41	-0.36
	School	Post_20	-0.34	-0.39

There was a pre-post decline in sCort concentration during Day 2 (Music), but this effect was not replicated on Day 4 (Music) where a small pre-post increase was observed. Moreover, a similar pre-post decrease was observed during Day 1 (No Music). H2 is therefore not supported by Harry's data. His results are also inconsistent

with H3 because his sAA activity showed pre-post increases in both Day 2 (Music) and Day 4 (Music) but no pre-post increases in either Day 1 (No Music) or Day 3 (No Music) (See Figures 6.16 and 6.17). At school, there were no consistent patterns in sCort concentrations or sAA activity across treatment days.

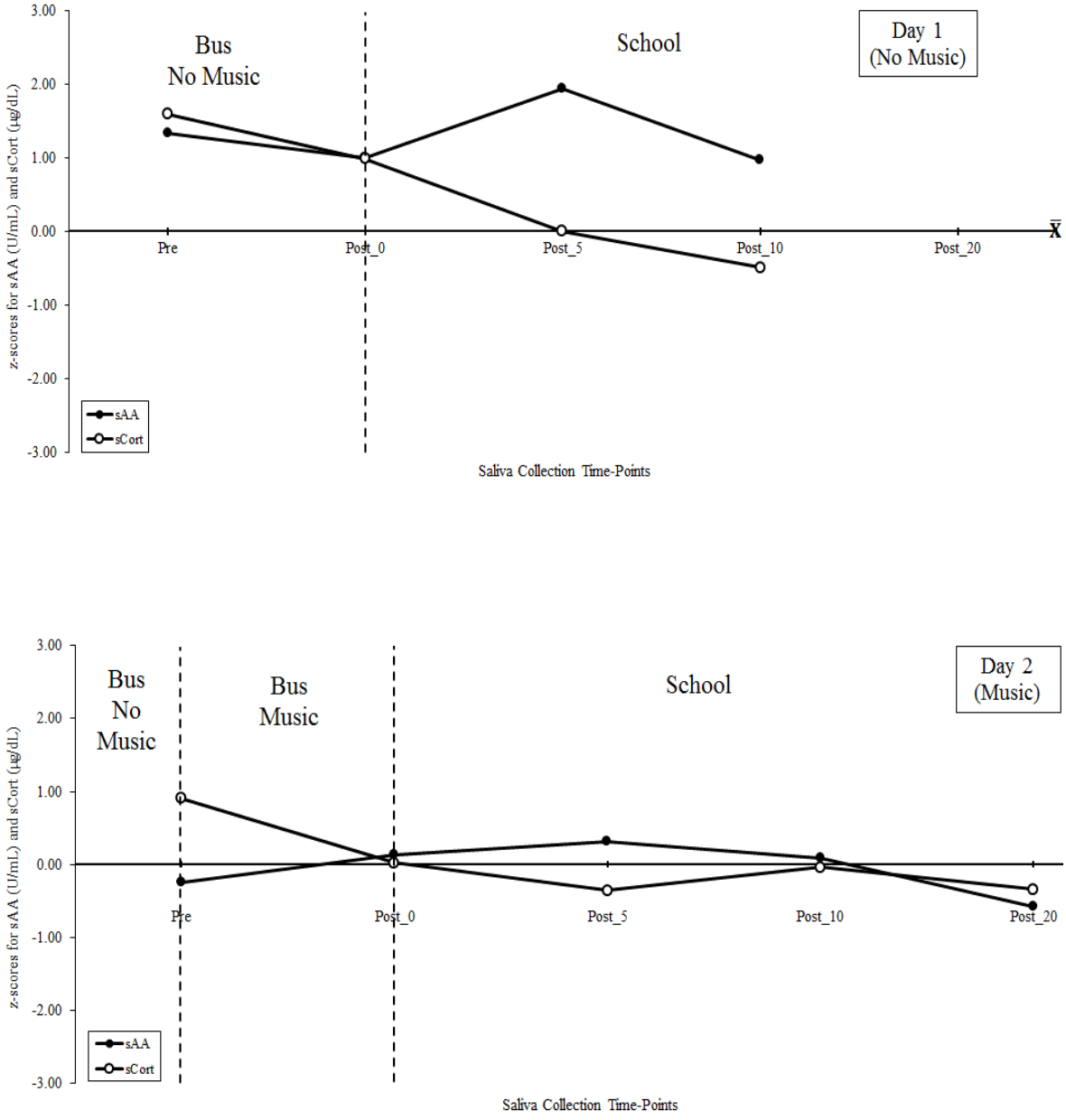


Figure 6.16 Harry's standardised sAA activity (U/mL) and sCort concentrations (µg/dL) (*y axis*) with saliva collection time-points (*x axis*) Day 1 (No Music) and Day 2 (Music) of Study 3.

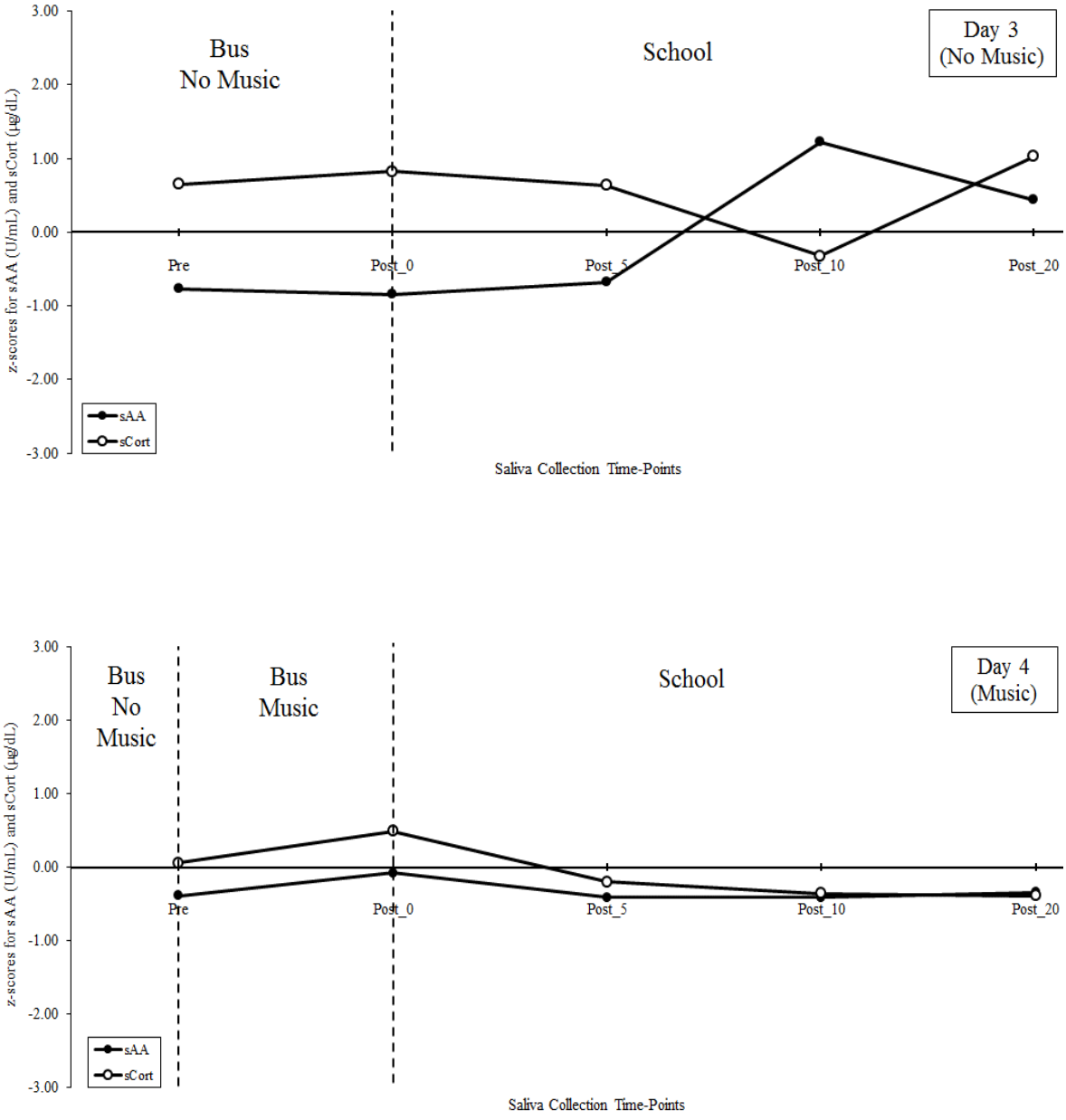


Figure 6.17 Harry's standardised sAA activity (U/mL) and sCort concentrations (µg/dL) (*y axis*) with saliva collection time-points (*x axis*) Day 3 (No Music) and Day 4 (Music) of Study 3.

6.6.11.5 SIB, sCort and sAA

H4 (reductions in sCort concentrations will be associated with reductions in SIB frequencies), H5 (reductions in sAA activity will be associated with reductions in SIB frequencies), H6 (changes in sCort concentrations will be associated with changes in sAA activity) and H7 (increases in SIB will be associated with either a symmetrical association or an asymmetrical association between sCort concentrations and sAA activity) were each tested with the Fisher's Exact test according to the procedure outlined in Section 6.4.2.

Contrary to H4, there was no association between sCort concentrations (above the mean level, below the mean level) and SIB frequencies (0%, between 0 and 100%, 100%) across the four treatment days of the study (p = .602). The 2 x 3 cell counts and frequencies are reported in Appendix U. Contrary to H5, there was no association between sAA activity and SIB frequencies across the four treatment days of the study (p = 1.000). The 2 x 3 cell counts and frequencies are reported in Appendix U. Contrary to H6, there was no association between sCort concentrations and sAA activity across the four treatment days of the study (p = 1.000). The 2 x 2 cell counts and frequencies are reported in Appendix U. Finally, contrary to H7, there was no association between joint sCort concentrations/sAA activity and SIB frequencies across the four treatment days of the study (p = .974). The 3 x 3 cell counts and frequencies are reported in Appendix U. Hypotheses 4 to 7 were therefore not supported by Harry's data.

6.6.12 Discussion

Harry's ear-blocking SIB was chosen as a target behaviour for the MAS by his primary carers. His MAS results indicated that his ear-blocking SIB was motivated by both sensory and tangible functions. Harry's primary carers suggested that his ear-blocking SIB may have been motivated by attempts to regulate and/or avoid auditory sensory stimuli. As such, reports of his refusal to wear protective ear muffs may have served several purposes. Firstly, despite being potentially effective in regulating and/or avoiding auditory sensory stimuli, wearing ear muffs may have made it difficult for Harry to continue ear-blocking as his established method. His insistence on ear-blocking and refusal to wear ear muffs may have also been impacted by cognitive deficits associated with his ID or inflexibility/rigidity associated with his ASD. Secondly, Harry's carers reported that his ear-blocking SIB became more intense when exposed to loud and unpredictable auditory stimuli such as barking dogs and the operation of household power tools. Ultimately, the function of Harry's ear-blocking

would have been best determined by a comprehensive FA prior to participation. In the absence of a FA, it is entirely possible that Harry's ear-blocking SIB served a different function all together.

Regarding tangible ear-blocking SIB functions, Harry's primary carers reported that he was rewarded with tangible food stuffs which were high in sugar upon task compliance until he reached 10 years of age. Hence, it is possible that Harry established a similar contingency to his ear-blocking SIB. For example, he may have ear-blocked or not ear-blocked dependent on whether a tangible reinforcer was provided. Again, a comprehensive FA would have provided valuable insights into the function of Harry's ear-blocking SIB and subsequent treatment administered in Study 3.

Harry's sCort concentrations were not found to be associated with his sAA activity across the saliva collection time-points. This result does not support the proposal that sCort concentrations and sAA activity are similar biomarkers of arousal (Bauer et al., 2002; Chrousos & Gold, 1992; Rudolph et al., 2010). However, Harry's results do support the finding that significant sCort concentration variance can be indicative of within group variation due to heterogeneity amongst children with ASD (Corbett et al., 2012).

Prior to the commencement of Study 3, Harry's primary carers believed that he would not comply with the saliva collection protocol due to oral sensory aversions. An example of this was Harry's refusal to take prescribed oral medications. If oral sensory aversions were a clinical concern for Harry, it may have influenced his sCort concentrations and sAA activity. Such considerations appear to be indicative of the complex and heterogeneous nature of ASD.

Regarding sAA, the existing body of knowledge about this biomarker amongst children with ASD is either non-existent or not yet sufficiently validated (Kidd et al., 2012; Quas, 2011). As such, Harry's non-significant sAA response to music listening and lack of correlation with the more empirically established sCort concentrations may be of value to the body of knowledge.

Overall, Harry's sCort concentrations and sAA activity were not found to be associated with his ear-blocking SIB frequencies. Similar to Bill, this finding does not support Jennett et al. (2011) who reported that an increased rate of SIB was associated with elevated levels of arousal. However, similar to what has previously been discussed in relation to Bill, this result may be useful for inclusion into an emergent

body of knowledge regarding biomarkers of arousal and SIB amongst children with LFA.

Harry's results provide no support for the assertions of either the additive or the interactive models published by Bauer et al. (2002). However, as previously discussed in relation to Bill's ear-blocking SIB, Harry's results may have been influenced by auditory sensory aversions or tactile sensory contingencies an association between music listening and ear-blocking SIB frequencies was not mediated by biological arousal. Overall, Harry's results matched those of Bill in that they failed to validate H1 to H7 and the theoretical three-stage mediating model.

6.7 Single-case presentation: George

The information contained in the following case presentation was gathered from the clinical research interview administered by the researcher to George's biological mother. To retain George's anonymity, his name has been altered. The content of his case was endorsed by his primary carer via a member checking procedure.

6.7.1 Family and developmental history

George was born weighing 4.1 kilograms. He lived with his biological mother and brother in a semi-rural suburb. George's mother held a Bachelor's degree in Biological Science and postgraduate qualification in school teaching. George's biological father had achieved a high school level of education. George's biological mother and father separated not long before he was born.

George's mother reported the first trimester of her pregnancy to be unremarkable apart from severe bouts of morning sickness. George's birthing process was complicated by meconium in the amniotic fluid, his shoulder was wedged in his mother's pelvis, with an unsuccessful suction before forceps delivery. As a result, George was born with an irregular shaped cranium, fractured left clavicle and lacking oxygen. His one-minute Apgar score was critically low before rising to a typical range at the 5-minute re-test.

George spent one week in an intensive care unit where he was given antibiotics for suspected meningitis and fed a mixture of his mother's milk and formula via nasogastric tube. His ability to accept his mother's breast was limited by an apparent oral sensory aversion and facial swelling associated with his complicated birth.

When he was discharged from hospital, George contracted colic, cried uncontrollably and was extremely difficult to soothe. Interestingly, he was only

soothed by very loud household machinery such as vacuum cleaners and by vigorous whole body movements such as bouncing on trampolines or being swung from his feet whilst upside down.

When breastfed, George's latch was excessively strong. This resulted in his mother resorting to bottle-feeding. He consistently bit through bottle nipples after very short periods of time. He also developed an excessive appetite at this stage. His mother had to feed him a mixture of expressed breast milk and formula in high volumes to satisfy his appetite.

George sat upright at 6 months, stood at 10 months and walked at 11 months. As a toddler, George climbed on familiar and unfamiliar people as if they were climbing frames, often acted with social inappropriateness, did not seek affection from others and was highly anxious when separated from his mother. Further, George repetitively pulled plugs in an out of electrical sockets, flicked light switches on and off, and opened and closed screen doors. Ritualistically, George became extremely distressed when the nozzle of the dish washing detergent bottle was not closed.

By 22 months of age, George completed shape sorting games and jigsaw puzzles with remarkable speed and accuracy, unscrewed toothpaste caps and brushed his own teeth. George was diagnosed with ASD at the age of four.

At 8 years of age, risk taking behaviours and an absence of fear were apparent. George climbed on and off high structures and stood on the kitchen bench to pull light globes from inside ceiling down lights. At this age, he also became more anxious and combative. His behavioural lability ranged from bouncing happily on the trampoline to laying on the floor, crying uncontrollably before engaging in pica SIB and hand biting. George's mother often woke during the night to find him sobbing uncontrollably and find him impossible to soothe. His medical practitioners suggested that his anxiety and aggression were the result of pre-pubescent hormones.

George underwent speech, occupational and behavioural therapy. However, the efficacy of these therapies was limited by his tiredness due to disturbed sleep patterns and refusal to re-visit therapy venues more than once. No indication of childhood illness, brain injury or infection was reported by George's mother.

6.7.2 Accommodation and schooling

George attended mainstream school until he was 6 years of age. By the age of 8, he had been transitioned to a special education unit within a mainstream school due

to non-compliance and other challenging behaviours. He was then transitioned to a special education school.

At school, George's classroom participation was consistently interrupted by his pica SIB. If school staff removed a pica object from his mouth, he soon found a replacement. Attempts to occupy his hands as a method of reducing access to pica objects were also unsuccessful. At home and school, George required constant support.

6.7.3 Physical attributes and demeanour

Fourteen year-old George weighed 62 kg and was 175cm tall at the time of the interview. He appeared to be physically strong with long and wavy blonde hair. His calf and core stabilising muscles were overly developed due to toe-walking and repetitively lunging forward and backward. The muscles along his jawline were also overly developed as a result of his excessive pica SIB.

George leant toward his left when standing and had a severely limited vision out of his left eye. As a result, he rotated his head to the left to make use of his unaffected right eye. George refused to wear glasses. George's mother recalled numerous occasions when he had bumped into immovable objects due to his visual limitation. On route from the school bus to classroom, George tapped metal posts and other objects to listen to the sound of the vibration.

6.7.4 Communication

Up until 3 years of age, George had use of approximately 20 words. However, not long after this, he progressively lost use of these until they disappeared altogether. George had no discernible verbal or non-verbal communication. When extremely excited or agitated, George produced a high pitched and loud squealing sound. At other times, he produced guttural groans and grunts. George's main method of communicating was through non-verbal gesture. For example, he led people to desired objects/activities by the hand and showed that he was pleased by hugging. In addition, he inconsistently used a picture exchange system to have personal care needs met.

George's facial expressions did not often match his mood. For example, he would smile when unhappy and frown when happy. His facial expressions were limited in number to happy, sad and flat. His eye contact was fleeting and avoidant.

6.7.5 Nutrition

George preferred to eat foods that produced audible crunching sounds such as corn chips and rice crackers. He also ate foods that required minimal chewing such as spaghetti, banana, yoghurt and ice cream. He consumed all food or drink at a fast rate.

6.7.6 Music

As an infant, George's mother sang him slow tempo nursery rhymes and played recordings of classical music to calm him when agitated. When he heard this music, George became physically still, orientated himself toward his mother's face or the speaker, relaxed and eventually fell asleep.

George listened to age-appropriate music as well as music for young children. He also played with saxophone and piano toys which produced childhood instrumental music repetitively. When his mother played him a recording of the third movement of Ludwig van Beethoven's Sonata No. 14 in C-sharp minor, Op. 27 (fast tempo), George spun in circles, squealed, ran uncontrollably throughout the house and increased the frequency and intensity of his pica SIB.

6.7.7 Behaviour

George's primary behavioural focus was to engage in pica SIB. Initially, his pica was coupled with a low toned guttural sound. However, after a continuous period of pica, he produced loud and high pitched squealing vocalisations. If his squealing did not abate, George would bite the back of his hands or hit himself in the forehead with objects such as plastic chairs or pieces of wood. This aggressive and self-injurious behavioural trajectory became more frequent since his risperidone medication had been reduced.

Day to day, George was not able to care for himself. For example, he did not use the toilet unassisted. Rather, George urinated and defecated behind chairs at home, in gardens at school and on the school bus. As a preventative measure, he wore incontinence pads daily.

George did not have friendships with same-aged peers. His social approaches appeared awkward and inappropriate. For example, he approached unfamiliar people with both hands outstretched before attempting to physically move them toward a desired/fixated object or activity. As such, George required intensive social supervision. When frustrated in social settings, George became angry and aggressive toward others.

George showed numerous repetitive behaviours. For example, he repetitively placed his hands down the front of his pants, rocked back and forth whilst standing and/or seated, and postured with his hands whilst inspecting them visually at close proximity. When George engaged in long periods of repetitive behaviour, he became dysregulated and physically aggressive toward himself and others.

6.7.8 Behavioural interventions and medications

Previous behavioural and pharmacological interventions designed to treat George's pica SIB had been ineffective. As a result, George's mother resorted to providing non-toxic and soft textured pica objects which were both safe to chew and minimised dental trauma. George wore a belt around his waist and lanyard around his neck with these pica objects attached. He also engaged in pica with a free chewing tube and plastic coil.

6.7.9 DSM-5 ASD features

George's mother described him as living in "a world of his own" where outcomes must go in his favour. Regarding the first domain of the DSM-5 diagnostic criteria for ASD, George had severe social difficulties in communication and interaction across all contexts, reciprocity, conversation, initiation, response, eye contact, body language, developing, understanding and maintaining relationships, use of communicative gestures and making friends. Further, he expressed a restricted range of emotions and facial expressions, lack of interest in same-aged peers and ability to adapt his behaviour to meet the requirements of social situations.

Regarding the second domain of the DSM-5 diagnostic criteria for ASD, George showed perseverative, restricted, repetitive, pervasive and predictable behaviour such as pica SIB. In addition, he lunged backwards and forwards vigorously, bounced on the spot in a self-stimulatory fashion and swayed from side to side. Further, he was drawn toward excessively loud sounds made by motorised garden tools and household machines. These sounds made him extremely excited then aggressive. His acceptance of these sounds was based on him being able to activate or cease the machinery making these sounds. For example, he enjoyed operating exhaust fans but was avoidant of the sound produced by automated hand dryers in restrooms.

George preferred not to wear shoes and was not perturbed by walking on sharp objects with bare feet. In the absence of formal assessments, George's mother suggested that he had an intellectual disability and severe anxiety. In addition, George exhibited persistent pica SIB and required support to safely manage daily living. He was often non-compliant and was delayed in response. Despite being aware of others, George preferred not to partake in social interactions unless it was instrumental to having an immediate need met. George possessed an acute spatial awareness. For example, at school and on the school bus, George lunged and jumped vigorously without coming into contact with others.

The above-mentioned features severely limited George's social and academic development at home, school and in the community. He accessed out-of-home respite care which positively impacted on the mental well-being of his mother and brother.

6.7.10 SIB: Pica

George's mother chose pica SIB as the target for the present research. This type of SIB occurs in approximately 36% of people with LFA (Lai et al., 2014). Pica is defined as placing an inedible and non-nutritional object onto the lips or inside the mouth (Singh & Winton, 1984). George's mother described his pica SIB as the persistent chewing of inedible objects which occurred across all environments and was often made worse by high anxiety. When engaged in pica for too long, George became dysregulated and aggressive toward himself and others.

George's pica SIB first emerged at 18 months of age as he began to bite through pacifiers and teething rings. As a toddler, he constantly chewed toy trains. As he grew, George progressed to chewing cardboard and hard plastic puzzle pieces, the covers of hard cover books, Lego blocks, gold jewellery, DVD covers, and USB thumb drives.

George's pica SIB became excessive at 8 years of age after a 12-month gluten and dairy free diet. His mother enforced this diet because she believed that foods containing gluten and dairy resulted in challenging behaviours. For example, George became dysregulated and aggressive after eating rice crackers. This diet was discontinued after all foods but buckwheat pancakes and natural pears had been eliminated and George's challenging behaviours had not abated.

At the age of 8 years, when his dietary restrictions were lifted, George chose to eat previously eliminated foods only. He ate one type of food exclusively for periods of time before moving to another food type. For example, he ate yoghurt for 7 days before replacing this with apples. During the food elimination and reintroduction, George's pica SIB became more frequent. Further, George's preference for audibly crunchy foods such as breakfast biscuits, cracker biscuits and rice cakes strengthened.

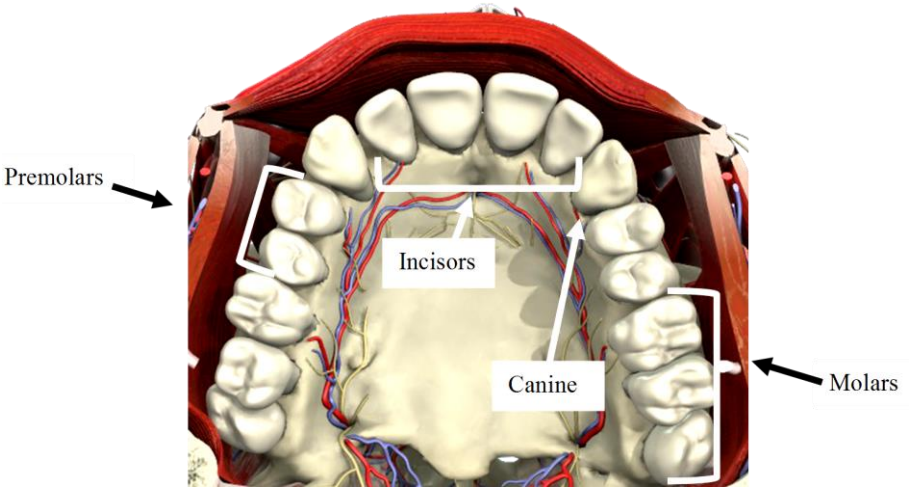
George's pica occurred consistently and pervasively for 7 years. Over this time, he had chewed on and eaten hair shampoo and glue bottles, chalk, play putty, marker pens, plastic handle bars, foam from couches, batteries, hard plastic game pieces, television remote controls, wooden objects and spectacles. George attended hospital numerous times for the removal of inedible objects.

George's preference for hard and inedible pica SIB objects resulted in damage to the structure and alignment of his molars, premolars, canines and incisors (see

Figure 6.18). George required substantial dental work, however he refused to attend the dentist. His jagged teeth created deep cuts within his mouth and on his hands when bitten. These injuries often resulted in infections and significant scaring. Further, the excessive saliva produced by his pica resulted in skin deep cracks on his fingers. His oral hygiene was poor due to him either refusing to have his teeth brushed or treating tooth brushes as pica objects.

Figure 6.18

Roof of oral cavity from below (Primal Pictures, 2006a).



George's family and carers monitored his behaviour at all times to ensure that he did not engage in pica with harmful objects/substances. To minimise possible harm, toxic and dangerous pica objects were hidden and he was rarely taken out into public where harmful pica objects were accessible. His family and carers were often anxious about the possibility of George choking or being poisoned as a result of his pica SIB.

George's pica SIB increased in frequency when anxious and reduced when engaged in favoured classroom activities such as jigsaw puzzles. However, his mother indicated that his pica SIB could function to calm or stimulate.

6.7.11 Results

6.7.11.1 SIB function

The MAS was administered to George's primary carer to determine if his pica SIB on the school bus and at school was motivated by attention, tangible objects, avoidance or sensory functions (see Appendix Y; Joosten & Bundy, 2010). An analysis of his MAS data ranked his pica SIB as being mostly motivated by sensory and tangible factors with some data to suggest escape and attention domain motivations (see Figure 6.19).

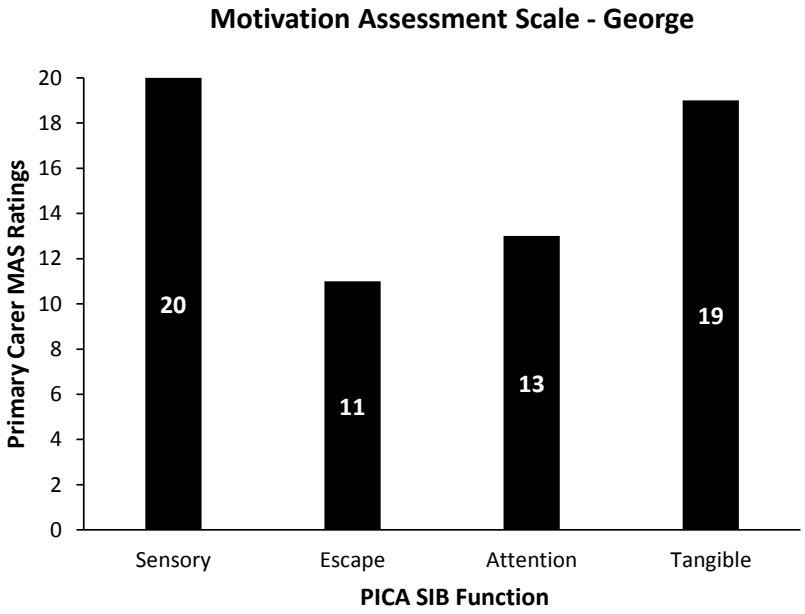


Figure 6.19 Study 3: George's MAS results.

6.7.11.2 Inter-rater agreement

Inter-rater agreement for footage recorded on the school bus and at school was assessed by the researcher and two independent and qualified raters as it was for Bill and Harry (Rater A and Rater B; see Appendix X). The number of 10-second intervals within which George exhibited pica SIB per minute were rated across each of the four treatment days. As seen in Table 6.6, inter-rater reliability was high across all three raters. Therefore, the interval ratings recorded by just one rater (Rater A) were used in the analysis.

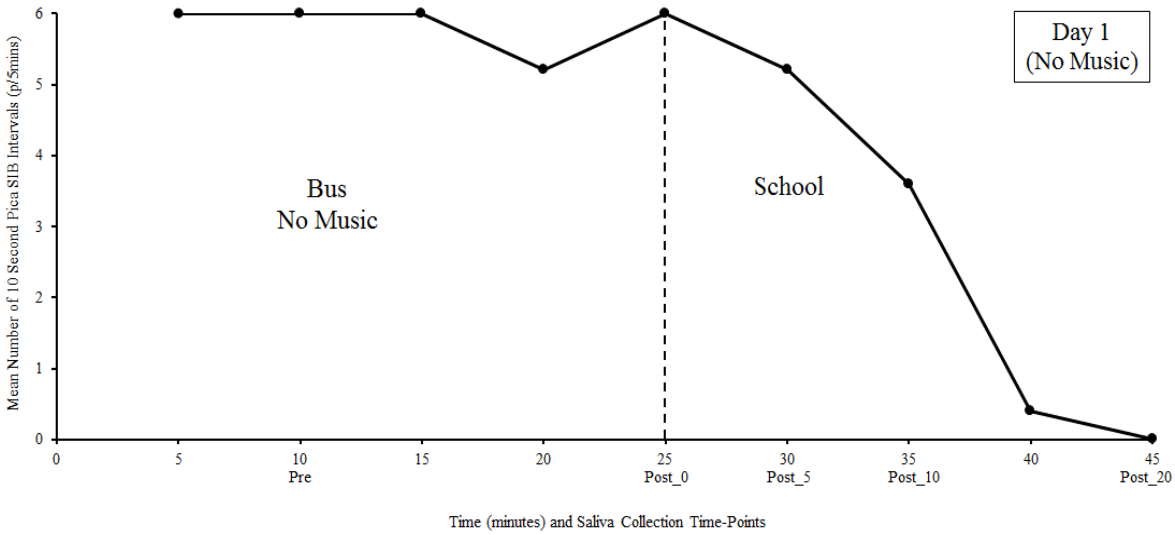
Table 6.6

Study 3: George's Inter-rater Agreement

Treatment Days	Inter-rater Agreement (the proportion of 10-second SIB interval ratings for which all inter-raters agreed)
Day 1: No Music	97.1% (201/207)
Day 2: Music	97.5% (192/197)
Day 3: No Music	98.1% (203/207)
Day 4: Music	96.3% (155/161)
Mean:	97.2%

6.7.11.3 Music listening and SIB

H1 (listening to calming music will reduce the frequency of SIB compared to non-exposure) was tested via visual inspections of Figures 6.19 and 6.20. On Day 2 (Music), the frequency of George's pica SIB dropped to zero shortly after the introduction of the music, and essentially stayed there for the duration of the music. This is in sharp contrast to both Day 1 (No Music) and Day 3 (No Music) where his pica SIB remained at relatively high levels throughout the bus journey. Although these data support H1, the support is tempered by the fact that the music effect did not replicate as seen on Day 4 (Music). Therefore, listening to calming music did not reduce George's pica SIB interval ratings during the Day 4 treatment. In addition, at school, no consistent SIB trends emerged on music or no music treatment days.



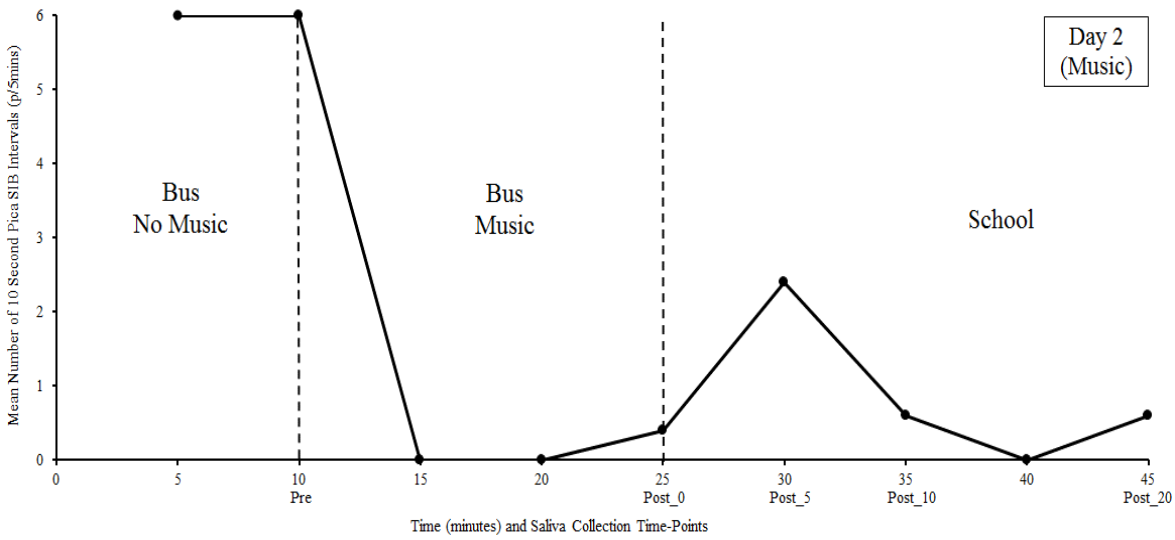
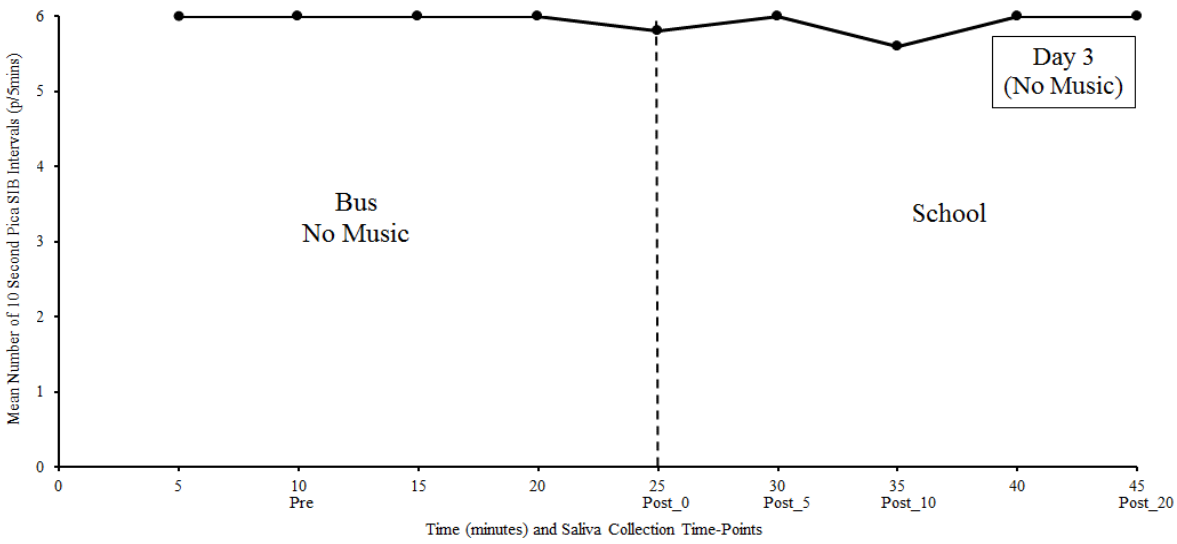


Figure 6.20 George's mean pica SIB intervals (*y axis*) with saliva collection time-points (*x axis*) for Day 1 (No Music) and Day 2 (Music) of Study 3.



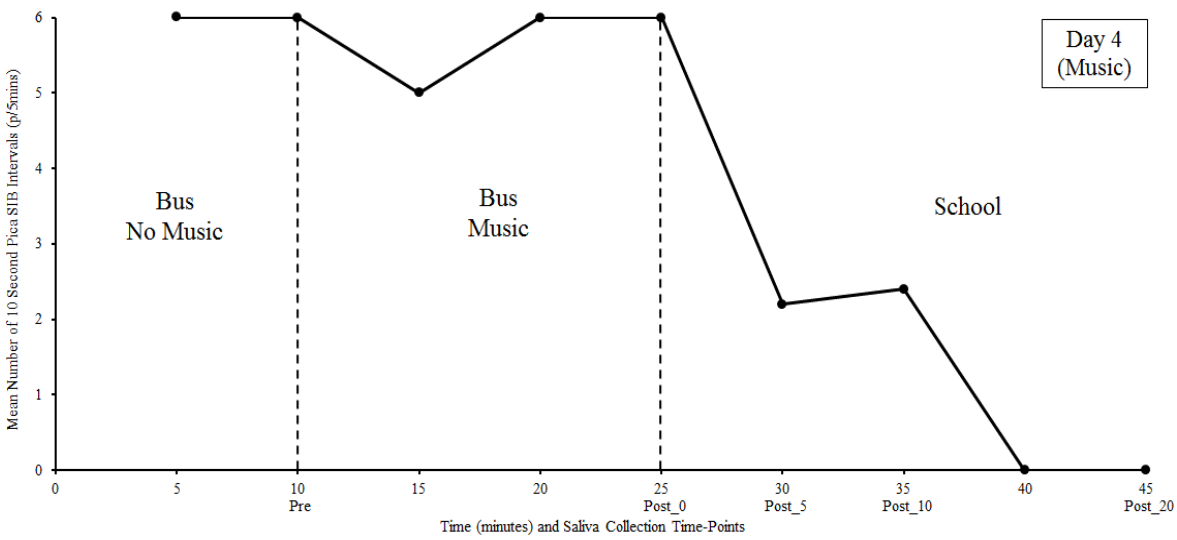


Figure 6.21 George's mean pica SIB intervals (*y axis*) with saliva collection time-points (*x axis*) for Day 3 (No Music) and Day 4 (Music) of Study 3.

6.7.11.4 Music listening, sCort and sAA

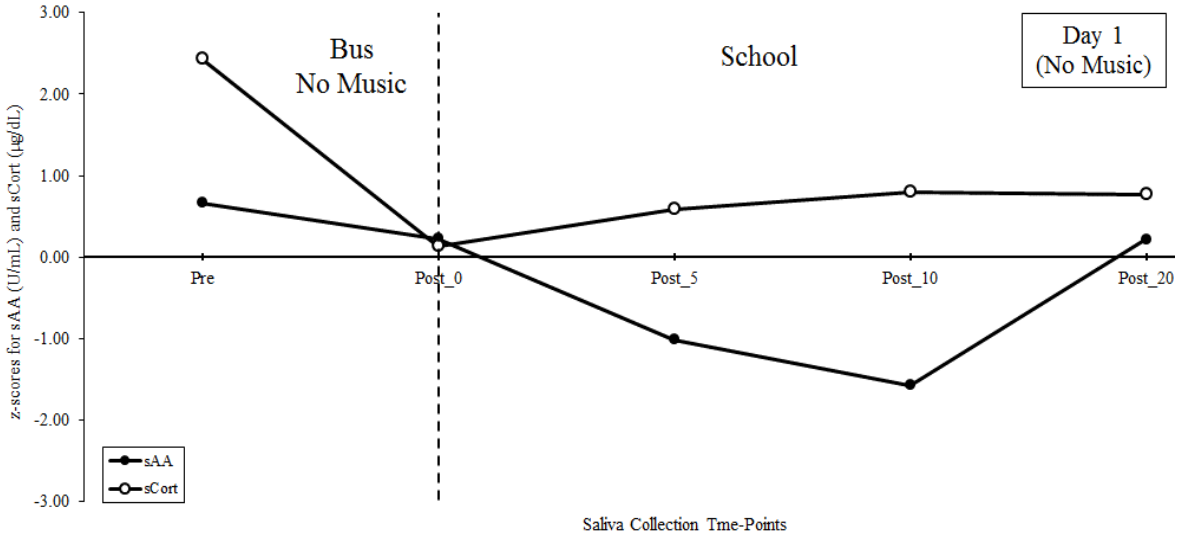
For H2 (listening to calming music will be associated with a reduction sCort concentrations compared to non-exposure) and H3 (listening to calming music will be associated with a reduction sAA activity compared to non-exposure), like Bill and Harry, the analysis of George's sCort concentrations, sAA activity and joint sCort/sAA concentration/activity were restricted to the standardised sCort and sAA measures taken at the Pre and Post_0 saliva collection time-points. As such, the following results should be viewed as suggestive only. The standardised scores are reported in Table 6.7.

Table 6.7

George's Standardised sAA Activity and sCort Concentrations Across the Five Saliva Collection Time-points for the Four Days of the Study

Treatment Day	Condition	Time-Point	sAA (U/mL)	sCort (µg/dL)
Day 1 (No Music)	Bus No Music	Pre	0.67	2.44
	Bus No Music	Post_0	0.23	0.13
	School	Post_5	-1.02	0.59
	School	Post_10	-1.57	0.80
	School	Post_20	0.22	0.77
Day 2 (Music)	Bus No Music	Pre	-1.00	-1.11
	Bus Music	Post_0	-0.51	-0.92
	School	Post_5	-0.92	-1.07
	School	Post_10	2.16	-1.05
	School	Post_20	-0.97	-0.44
Day 3 (No Music)	Bus No Music	Pre	0.63	-2.02
	Bus No Music	Post_0	-0.37	0.70
	School	Post_5	-0.08	0.28
	School	Post_10	0.11	0.61
	School	Post_20	-0.52	-0.17
Day 4 (Music)	Bus No Music	Pre	1.62	-0.68
	Bus Music	Post_0	1.61	-0.69
	School	Post_5	0.21	-0.34
	School	Post_10	0.47	-1.05
	School	Post_20	-0.96	-0.64

As seen in Figures 6.22 and 6.23, there were negligible pre-post changes in sCort concentrations or sAA activity during Day 2 (Music). There appeared to be an increase in sCort concentrations during Day 3 (No Music), but this wasn't replicated during Day 1 (Music) where a reduction occurred. At school, there was no consistent patterns in sCort concentrations or sAA activity across treatment days.



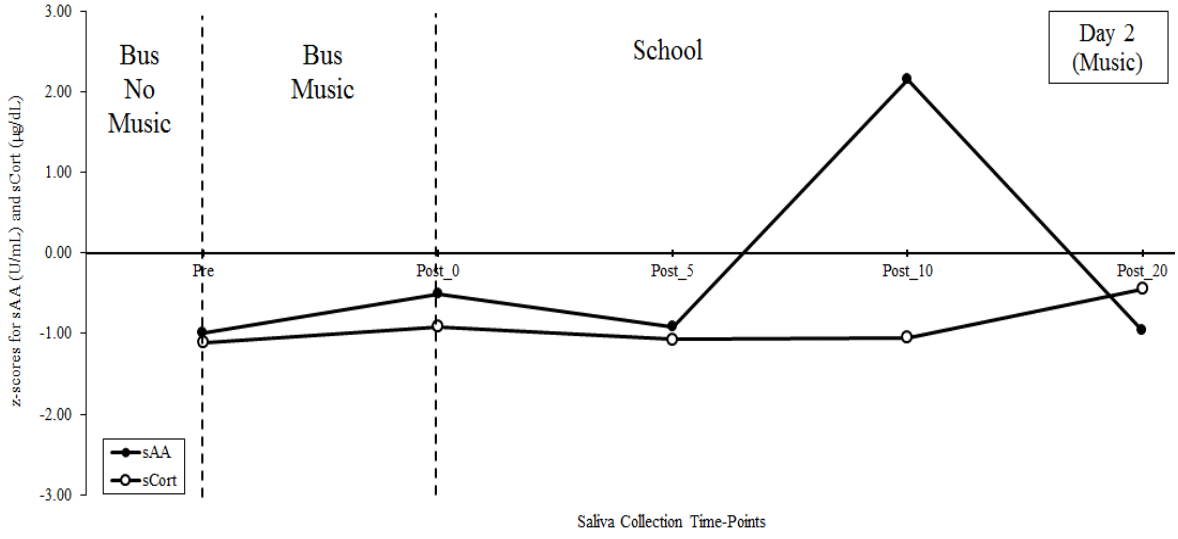


Figure 6.22 George's standardised sAA activity (U/mL) and sCort concentrations (µg/dL) (*y axis*) with saliva collection time-points (*x axis*) Day 1 (No Music) and Day 2 (Music) of Study 3.

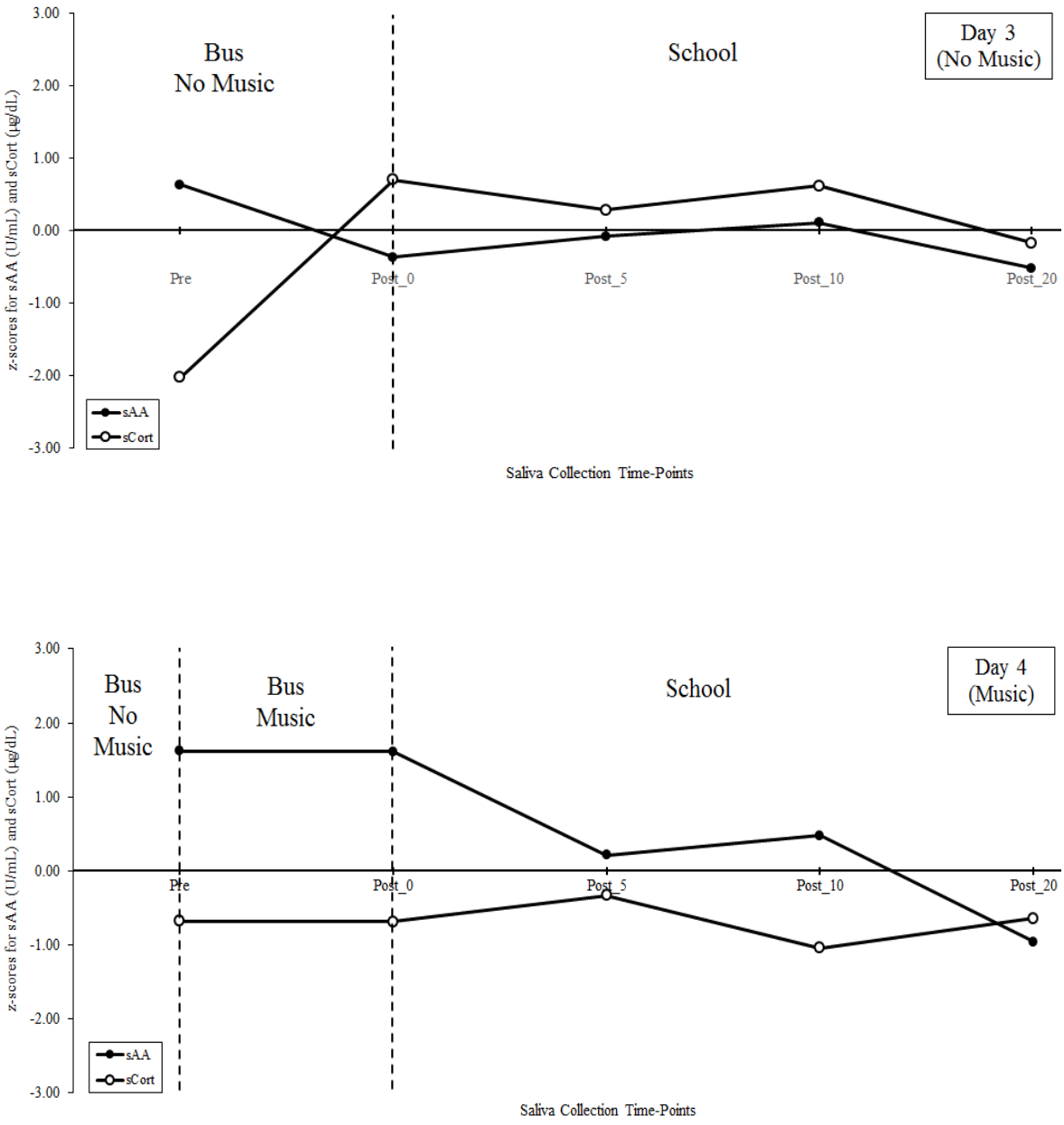


Figure 6.23 George's standardised sAA activity (U/mL) and sCort concentrations (µg/dL) (*y axis*) with saliva collection time-points (*x axis*) Day 3 (No Music) and Day 4 (Music) of Study 3.

6.7.11.5 SIB, sCort and sAA

H4 (reductions in sCort concentrations will be associated with reductions in SIB frequencies), H5 (reductions in sAA activity will be associated with reductions in SIB frequencies), H6 (changes in sCort concentrations will be associated with changes in sAA activity) and H7 (increases in SIB will be associated with either a symmetrical association or an asymmetrical association between sCort concentrations and sAA activity) were each tested with the Fisher's Exact test according to the procedure outlined in Section 6.4.2.

Contrary to H4, there was no association between sCort concentrations (above the mean level, below the mean level) and pica SIB frequency (0%, between 0 and 100%, 100%) across the four treatment days of the study (p = .377). The 2 x 3 cell counts and frequencies are reported in Appendix U. Contrary to H5, there was no association between sAA activity and pica SIB frequencies across the four treatment days of the study (p = .637). The 2 x 3 cell counts and frequencies are reported in Appendix U. Contrary to H6, there was no association between sCort concentrations and sAA activity across the four treatment days of the study (p = 1.000). The 2 x 2 cell counts and frequencies are reported in Appendix T. Finally, contrary to H7, there was no association between joint sCort concentrations/sAA activity and SIB frequencies across the four treatment days of the study (p = .477). The 3 x 3 cell counts and frequencies are reported in Appendix U. Hypotheses 4 to 7 were therefore not supported by George's data.

6.7.12 Discussion

Pica SIB was chosen as the target behaviour for the MAS and treatment by George's mother as a result of associated physical injuries and safety concerns. Results of the MAS indicated that George's pica SIB was mostly motivated by sensory and tangible factors. In relation to the sensory factor, the MAS does not indicate which specific sensory factor motivates George's SIB. However, George's mother reported that his pica SIB appeared to be motivated by an oral sensory seeking tendency. The longer George engaged in pica, the more likely he was to become over-aroused and injurious toward himself and others. Further, he was known to attempt to place entire pica SIB objects into his mouth. This appeared to result in full mouth sensory input. An indication of his oral sensory seeking for his early childhood may have also been

his excessively strong latch whilst being breast and bottle fed. In addition to this evidence, a FA would have been beneficial in accurately determining the function of George's pica SIB.

George's sCort concentrations were not associated with his sAA activity across the saliva collection time-points. This did not support the hypothesis that sCort and sAA can both be viewed as indicators of a homeostatic response (Bauer et al., 2002; Chrousos & Gold, 1992; Rudolph et al., 2010). Considering that the same result was identified across Bill and Harry's data, it reinforces the validity of the Corbett et al. (2012) paper which reported significant variance within the sCort concentrations amongst children with ASD due to heterogeneity. Further, it reinforces the need to study sAA activity to determine its validity as a biomarker of arousal for children with ASD. As previously discussed regarding Bill and Harry, despite the lack of correlation between sCort concentrations and sAA activity in this study, these individual and now collective results may be meaningful to the body of knowledge regarding the function of the HPA axis and ANS amongst children with LFA.

George's sCort concentrations and sAA activity were not associated with his pica SIB frequencies. As previously discussed, this result does not support the Jennett et al. (2011) paper which reported that an increased rate of SIB can be linked to elevated levels of arousal.

Similar to the results of Bill and Harry, a combination of George's results was used to assess the validity of the additive and the interactive models published by Bauer et al. (2002) assuming that the pica SIB can be seen as a challenging behaviour and that LFA is a profound disability. As seen in the results for Bill and Harry, the HPA axis as indicated by sCort concentration was not associated with the ANS as indicated by sAA activity across the saliva collection time-points. Further, no association was found between sCort concentrations or sAA activity. Pica SIB frequencies were found not to be associated with sCort concentrations or sAA activity. In sum, the above-mentioned results provide no support for the assertions of either the additive or the interactive models (Bauer et al., 2002). Moreover, the hypothesis that biological arousal may mediate a relationship between music listening and pica SIB was not supported; therein not supporting the theoretical three-stage mediating model proposed. However, as George's MAS results suggest, his pica SIB may have functioned to meet an oral sensory need. Therefore, despite the lack of sCort and sAA association, his biological data may still provide a meaningful contribution to the

emerging body of knowledge regarding the biological arousal of children with LFA who exhibit SIB.

During Day 2, when exposed to the *Sonata Pathetique*, George's pica SIB frequency dropped to zero and stayed there for its duration. Despite this result, the same music effect was not observed at the repeat treatment on Day 4. Hence, the music effect observed during Day 2 was likely the product of an extraneous factor.

6.8 Summary

The heterogeneity of LFA is apparent in its diagnostic variance, presentation, behaviour and now it seems, biologically determined arousal. On the back of statistically significant results found by listening to calming to music in the controlled school bus simulation environment of Study 2, it was hypothesised that music may again reduce sCort concentrations and potentially sAA activity during an actual and naturalistic morning school bus ride environment. If this were achieved, it may have been further tested that reducing sCort concentrations and sAA activity through music listening may have also reduced SIB frequencies. However, this was not the case for numerous reasons.

Firstly, associations between sCort and sAA, sCort and SIB, sAA and SIB were not detected. In combination, these results did not support the theoretical three-stage mediating model proposed. Moreover, it not only appears that high degrees of heterogeneity exist within groups of children with ASD as reported by Corbett et al. (2012), the results of the current research suggest that such heterogeneity is also apparent amongst the three participants of Study 3. Further, the inclusion of a comprehensive FA to augment the MAS may have determined the function, in addition to motivating factors, underpinning SIB exhibited by each participant. This functional analytic data may have influenced the choice of treatment, which may have influenced the sCort, sAA and SIB results. Lastly, single-case presentations such as George's have the potential to uncover the possible underpinnings of drastic behaviour change such as that seen during Day 2. For this to occur, further repetitions of the treatment would have been required.

CHAPTER 7: DISCUSSION AND CONCLUSION

Interest in the behaviour of children with ASD began with the seminal articles published by Leo Kanner and Hans Asperger in the 1940s (Asperger, 1944 as cited in Frith, 1991; Kanner, 1943). Within these articles, single-case presentations of Donald T. described as "extremely autistic" (Kanner, 1943, p. 222) and Fritz V. identified as "highly unusual" (Asperger, 1944 as cited in Frith, 1991, p. 39) perpetuated this interest. As a result, and ever since, research has been conducted with the aim of understanding more about ASD, its prevalence, potential causes, and how to treat the most challenging of its behaviours (Baker, 2013).

The most serious and frequently occurring of challenging behaviours exhibited by school-aged children with ASD is self-injurious behaviour (SIB; American Psychiatric Association, 2013; Horner et al., 2002; Matson & Nebel-Schwalm, 2007; McClintock et al., 2003; Minshawi, 2008). SIB ranges in severity from mild and occasional to severe and frequent (Fee & Matson, 1992). SIB is exhibited by up to 40% of people with an ID such as those with LFA (Bodfish et al., 1995; Kahng, Iwata, & Levin, 2002). If not treated effectively, challenging behaviours such as this can significantly impact on the physical, academic and social development of children with LFA (Glasson et al., 2008; Matson, Sipes, et al., 2011; McClintock et al., 2003).

Treating SIB exhibited by those with LFA can be complex and testing (Briere & Gil, 1998; Novak, 2003; Smith et al., 2005). Applied Behaviour Analysis (ABA) techniques have led to positive behavioural outcomes for treating such behaviours amongst people with ASD for over 40 years (Hastings & Noone, 2005; Matson & LoVullo, 2008; Matson et al., 2012). However, the ABA body of knowledge research could benefit from more publications in the area of treating self-aggressive behaviour such as SIB (Lai et al., 2014). When successfully implemented, pharmacological therapies for children with ASD who exhibit SIB can lead to greater therapeutic outcomes and school participation (Politte & McDougle, 2014), although the efficacy of pharmacological treatments for SIB, such as risperidone and aripiprazole, are often limited by medication resistance and adverse effects (Adler et al., 2015). Moreover, it has been suggested that the current behavioural and pharmacological treatments designed to treat repetitive behaviour such as SIB for children with ASD lack quantity and efficacy (Boyd et al., 2013).

People with ASD can respond positively to music (Papagiannopoulou, 2015). More specifically, music listening, defined as exposure to pre-recorded or live music, has been associated with favourable treatment outcomes for people with neurodevelopmental disorders such as LFA (Ockelford, 2013 as cited in Fancourt et al., 2014; Hooper et al., 2010). Of note, and as a potential treatment, music listening which satisfies an internal need such as reducing arousal, may minimise challenging behaviours such as SIB for those with limited communicative abilities such as LFA (Ford, 1999).

To assess the potential for music listening to reduce SIB, three studies were undertaken to determine the validity of a theoretical three-stage mediating model. Specifically, three separate but interlinked studies investigated the potential for biological arousal, as indicated by sCort concentrations and sAA activity, to mediate an association between listening to Beethoven's *Sonata Pathetique* and SIB among school-aged boys with LFA.

7.1 Key Findings

7.1.1 Study 1.

To account for the absence of a comprehensive music selection methodology in music-based intervention research, Study 1 incorporated expert musician consultations, period of musical composition, musical form and purposeful selection to identify a calming music playlist comprising six solo piano performance segments. The aim of Study 1 was to identify a single most calming musical segment for use in Study 2 and Study 3 through the application of a novel music selection methodology. The first 2 minute segments of Beethoven's *Moonlight Sonata* and *Sonata Pathetique* were rated by primary carers of males with LFA as significantly more calming. Subsequently, an expert musician consultant was re-engaged to make the final selection of Beethoven's *Sonata Pathetique* as the most calming musical segment.

The music selection methodology described in Study 1 may be useful in full or part thereof for future music-based research using music from the classical era, composed in Sonata form or to assess the impact of listening to other musical titles/forms. It may also improve replicability of music-based intervention research and validity of findings.

Amongst other conclusions made in the meta-analysis published by Cannella et al. (2005), it was reported that treatment preference can result in more favourable behavioural outcomes for the treatment of people with severe and profound

disabilities. However, the participants of Study 2 and Study 3 with LFA did not select their own music. This decision was enacted because it was believed that the ID component of the LFA may have resulted in difficulties for participants when attempting to operate the 7-point music selection Likert scale and comprehend the word *calming* for rating the six musical segments. When people with an ID such as those with LFA experience difficulties selecting music, their primary carers can help (Hooper et al., 2010). As such, it was deemed more appropriate for primary carers of boys with LFA to rate the music. It is noted that enabling boys with LFA to choose their own music may have been possible via a capability-based method, however measuring the accuracy of participants' ratings and comprehension of the word *calming* would have been required. The testing and establishment of such methods amongst those with LFA may provide a direction for future research.

7.1.2 Study 2.

The aim of Study 2 was to determine if listening to Beethoven's *Sonata Pathetique* could calm school-aged boys with LFA in a controlled environment. Specifically, Study 2 investigated if music listening resulted in reductions of sCort concentrations and sAA activity for boys with LFA after they were exposed to an adapted Trier Social Stress Test for Children (TSST-C) demand condition in the form of a morning school bus ride simulation. It was hypothesised that the sCort concentration and sAA activity of boys who listened to the *Sonata Pathetique* would decrease significantly compared to their matched controls. A significant decrease in sCort concentrations was detected amongst the group of boys who listened to the *Sonata Pathetique* whilst no decrease was detected in the control group. Significant post hoc decreases in sAA activity were also detected at several time-points amongst the music listening group whilst no decrease was detected amongst the controls. In relation to this result, it is possible that stronger sAA effects would have emerged with a larger sample size.

Future research into the validity of cortisol and alpha-amylase as biomarkers of arousal for children with ASD has been recently suggested (Bitsika et al., 2015; Bosch, Veerman, de Geus, & Proctor, 2011; Quas, 2011). As such, Study 2 detailed the saliva collection, and analysis of sCort concentrations and sAA activity amongst children with LFA. This addressed the need for the collection and analysis of salivary biomarkers of arousal as a non-invasive and socially appropriate method of the

measuring HPA axis and ANS function amongst people with special needs such as LFA (Davis & Granger, 2009; Fortunato et al., 2008; Gordis et al., 2006; Taylor et al., 2013).

7.1.3 Study 3.

Study 3 tested the validity of a theoretical three-stage mediating model investigating the potential for biological arousal (sCort concentrations and sAA activity) to mediate a relationship between music listening (exposure to Beethoven's *Sonata Pathetique*) and SIB frequencies. Data collected from the three participants with LFA revealed that sCort concentrations and sAA activity were not associated with reductions in SIB. As a result, the theoretical three-stage mediating model was not supported. This indicates that the biologically calming effect of listening to the *Sonata Pathetique* identified in the controlled environment of the school bus simulator of Study 2 did not generalise to the naturalistic setting of the actual school bus ride of Study 3. This may have been the result of uncontrolled extraneous variables which are discussed in section 7.5.

sCort concentrations and sAA activity did not correlate as biomarkers of arousal. As a result, the boy's data did not support either the additive model or interactive model proposed by Bauer et al. (2002). Despite the existence of published literature supporting the premise of both the additive and interactive models, the nature of an association between the HPA axis and ANS is mostly unknown. Some research even suggests that no association exists (Lisonbee, Pendry, Mize, & Gwynn, 2010; Spinrad et al., 2009). Recently, it was reported that the way in which the HPA axis and ANS interact, integrate and influence each other in neurotypical children is not yet categorically known (Koss et al., 2014).

It is noted that the results of Study 3, assessing the complete theoretical three-stage mediating model, may have produced different results with the inclusion of additional participants. Further, a moderator model may have been used as a more statistically robust method of assessing potential correlations between music listening, biological arousal and SIB. However, SIB, sCort and sAA data collected from at least 60 boys with LFA who exhibited SIB often would have been required to assess such a model.

7.2 Theoretical Implications

7.2.1 The additive and interactive models published by Bauer et al. (2002).

"We must show meaningful links between behaviour and physiology" (Kagan et al., 1994, p. 140). In pursuit of this, Bauer et al. (2002) published two theoretical

models; the additive model and interactive model. The additive model suggested that challenging behaviour such as SIB is more likely to be exhibited by children with developmental disabilities when there is a symmetrical association between HPA axis and ANS. Gordis et al. (2006) published support for the additive model based on sCort concentrations and sAA activity of adolescents before and after exposure to a modified story completion and mathematical subtraction TSST. Further, El-Sheikh et al. (2008) published support for the additive model based on the assessment of sCort concentrations and sAA activity amongst sixty-four 8 and 9-year-old children before and after a laboratory based exposure to a recorded argument between adults.

Study 3 found no associations between the HPA axis as indicated by sCort concentrations and the ANS via sAA activity. Hence, no support was identified for Bauer et al.'s (2002) additive or interactive models. This finding was further highlighted when no associations were found between music listening, sCort concentrations and sAA activity. Several factors may have contributed to these results such as undetermined functions of SIB due to the lack of Functional Analyses, the influence of uncontrolled variables in the naturalistic setting, natural variations in biological arousal and the heterogeneity of LFA. In addition, the limited number of participants in Study 3 yielded results of lower statistical power. As such, increasing the number of participants in Study 3 may have resulted in greater statistical power and enabled more advanced analyses. Further, assessing the possible effects of listening to different types of music or the effect of music listening on samples of neurotypical children on school buses as a preliminary investigation may have been more appropriate.

7.2.2 Rondo musical form calms boys with LFA.

In addition to selecting a single most calming segment of music for use in Study 2 and Study 3, Study 1 employed a novel music selection methodology. As a result, the second movement of Ludwig van Beethoven's *Sonata Pathetique* was selected. Not only did this musical segment provide an independent variable for use in Study 2 and Study 3, it selected music chosen that was composed in a particular form; the sonata form. Listening to the *Sonata Pathetique* in the controlled environment of Study 2 resulted in a significant decrease in sCort concentrations amongst school-aged boys with LFA when exposed to a simulated demand condition. Based on this result, music composed in sonata form appeared to be appropriate for use in music-based treatments designed to calm school-aged boys with LFA when exposed to demand conditions in

controlled environments. However, when the *Sonata Pathetique* was reduced to the first 2-minute segment of the second movement, it resembled a Rondo composition. Hence, it is suggested that music composed in rondo form may be more appropriate to calm school-aged boys with LFA as opposed to sonata. Such an assertion requires further testing.

With clinical relevance, the rondo musical form is identified by a repetitious and predictable structure (Abdy-Williams, 1890; Bennett, 1980; Randel, 2003). This structure appears to correlate with the "insistence on sameness" feature of the ASD component of the LFA diagnosis as described in the DSM-5 (American Psychiatric Association, 2013). However, it is entirely possible that other aspects of the *Sonata Pathetique* were the catalyst for the significant reduction in sCort observed in Study 2.

7.3 Clinical Implications

7.3.1 Music listening to reduce biological arousal in controlled versus naturalistic settings.

The results of Study 2 revealed that boys with LFA recorded a statistically significant reduction in sCort concentration as a result of listening to the *Sonata Pathetique* in the controlled environment of the morning school bus ride simulator. The simulated bus ride was chosen as a demand condition based on several ASD attributes as components of the LFA diagnosis. Firstly, people with ASD can be averse to changing environments such as the school bus due to "… extreme distress at small changes…" and "… need to take the same route…" (American Psychiatric Association, 2013, p. 50). Secondly, a "hyper- or hyporeactivity to sensory input or unusual interest in sensory aspects of the environment…" and "… adverse response to specific sounds or textures…" apparent within the school bus environment would likely create demands (American Psychiatric Association, 2013, p. 51). Having justified the school bus simulator as a demand condition, it may be assumed that the reductions in sCort concentrations recorded in Study 2 may occur for boys with LFA in other demanding but controlled environments such as the doctor's room, dentist's surgery or the therapist's room. Moreover, it is suggested that boys with LFA may benefit from services delivered in these environments when in a state of lower biological arousal.

Based on the results of Study 2, it may have been assumed that the significant reduction in sCort concentration would generalise to naturalistic settings such as the

actual school bus ride of Study 3. However, the results of Study 3 found no associations between music listening and sCort concentrations or sAA activity. This may have been due to uncontrolled factors occurring in the naturalistic setting of the school bus.

7.4 Research Strengths

Not detailing music selection methods in music-based intervention research can reduce the validity and generalisability of findings (Robb & Carpenter, 2009). Music selection methods are absent from meta-analyses, systematic reviews, critical examinations and a Cochrane review (Accordino, Comer, & Heller, 2007; Brown & Jellison, 2012; Gold, Voracek, & Wigram, 2004; Gold et al., 2006; Hooper, Wigram, Carson, & Lindsay, 2008; Simpson & Keen, 2011; Whipple, 2004). Further, research may not include music titles and/or the form in which music was composed (Lanovaz, Fletcher, & Rapp, 2009; Rapp, 2007; Stephens, 2008), the qualifications of music selectors (Carnahan, Basham, & Musti-Rao, 2009; Corbett, Shickman, & Ferrer, 2008; Simpson & Keen, 2010), include uncredentialed music raters, or limit music selection to pre-determined playlists (Crncec et al., 2006; El Hassan, McKeown, & Muller, 2009; Lesiuk, 2008). This appears to be a limitation of prior music-based interventions.

Regarding ASD music-based interventions, some authors have included credentialed musicians (Katagiri, 2009), primary carers (Pasiali, 2004), specific music titles and sources (Dellatan, 2003; Kern, Wakeford, & Aldridge, 2007; Orr, Myles, & Carlson, 1998) in the selection of music. Despite these, there is a current need for music-based interventions to meet minimum scientific standards and form a standardised method of music selection (Bradt, 2012; Robb & Carpenter, 2009).

Study 1 incorporated factors of calming music, specific musical, a music playlist selected by a concert pianist and primary carers. In doing so, this offered a novel method of selecting music for ASD music-based interventions that may be replicated in future studies.

Study 2 investigated the potential for music listening to reduce salivary biomarkers of arousal (sCort, sAA) among school-aged males with LFA both before and after a simulated morning school bus ride. The visual, audio and spatial dimensions of the simulator ensured realism, averted simulator sickness and formed a modified version of the TSST-C. Hence, the saliva sampled and data resulting offered novel contributions to the body of knowledge regarding biological responses to demand conditions in controlled environments for boys with LFA.

The RCT research design of Study 2 further improved the validity of the significant sCort reduction finding for the music listening group by minimising any participant selection bias. This result indicated that sCort concentrations and sAA activities derived from controlled experimental settings are valid measurements of biological arousal amongst boys with LFA.

Kagan said "We must show meaningful links between behaviour and physiology" (Kagan et al., 1994, p. 140). Prior to Study 3, previous research had not assessed both SIB behavioural and sCort with sAA data amongst boys with LFA. Although no associations were identified between SIB, the HPA axis as indicated with sCort, or the ANS as indicated by sAA, examining both behavioural and physiological markers was a strength of the study adding to our understanding of these associations.

Single-case research designs used to examine school-aged children with ASD are common in the field of psychology and can be both time-efficient and cost-effective (Lange & Lainhart, 2009; Machalicek et al., 2007; Yin, 2003). The information presented within the single-case presentations of Bill, Harry and George in Study 3 was the result of several research design components. Firstly, the primary carer clinical interview protocol administered to Bill, Harry and George's primary carers was based on empirical sources such as the seminal single-case reports of Donald T. and Fritz V. published by Leo Kanner (1943) and Hans Asperger (1944) respectively, the current DSM-5 diagnostic criteria and several peer-reviewed SIB single-case publications (American Psychiatric Association, 2013; Ford, 1999; Wachtel et al., 2011). To add further depth and detail to each case, the Motivation Assessment Scale (MAS) was administered to primary carers to present information about the potential motivations underpinning each participant's SIB. It is suggested that including the above-mentioned single-case design features strengthened the results and clinical relevance of Study 3 for primary carers, fellow researchers and those who work with boys who have who have a LFA. Further, this approach resulted in a least restrictive research method for the treatment of SIB (Hastings & Noone, 2005). Furthermore, greater numbers of participants and functional analyses, would have provided additional data and potentially different results.

7.5 Research Limitations

A key element of ABA research is the completion of a comprehensive Functional Analysis (FA) prior to commencement (Cooper, Heron, & Heward, 2007; Kennedy, 2005). Such assessments, based on information provided by primary carers

and school teachers, can identify a mode of treatment most likely to reduce SIB for the participants based on behavioural contingencies that increase or decrease the frequency of behaviours. As previously reported, conducting a comprehensive FA requires specific training and skills (Hastings & Noone, 2005). Unfortunately, the researcher did not possess the training or skills required to complete a FA at the time of the research. With similar intent, the MAS was used to identify the possible underpinnings of participants SIB in Study 3 (Durand & Crimmins, 1988). However, as previously noted, the reliability and validity of MAS is questionable (Koritsas & Iacono, 2013; Sigafoos, Kerr, & Roberts, 1994; Zarcone et al., 1991). As a result, the MAS results reported and integrated into Study 3 must be assessed with caution. In retrospect, a qualified behaviour analyst may have been sourced to complete a comprehensive FA for the three boys prior to the commencement of Study 3.

Sleep disturbances, physical sickness, daily routine and environmental changes, sensory aversions, communication difficulties, and the inability to express ritualistic and repetitive behaviours can trigger SIB for people with ASD (Adler et al., 2015; Boyd et al., 2013; Chicoine et al., 2013; Green et al., 2006; Lai et al., 2014; Matson & LoVullo, 2008). Further, people with more than one disability, such as those with LFA, are likely to exhibit SIB (Bright, Bittick, & Fleeman, 1981; Lai et al., 2014). In addition, SIB can be linked to behavioural contingencies such as social attention, social avoidance or operant functions such as sensory feedback (Barrera, Violo, & Graver, 2007; Iwata et al., 1994). Further, the occurrence of SIB is often the result of multiple factors that are specific to each individual (Adler et al., 2015). A primary method of determining the function of SIB, therein limiting the effect of extraneous variables, is to conduct a FA to identify if the function of SIB is attention seeking, obtaining a desired object, task avoidance or to obtaining a reinforcement (Hastings & Noone, 2005; Kahng et al., 2002; Posey et al., 2008). This method has a well-established peer-reviewed record and can detect environmental contingencies before informing treatment options (Beavers et al., 2013; Davis, 2015; Iwata et al., 1982; Iwata, Dorsey, et al., 1994). This is further justification for the usefulness of FA in the current research. It may have also been useful to augment the FA with the Questions About Behavioural Function (QABF) scale (Paclawskyj et al., 2000).

In relation to sCort, autoimmune disorders such as asthma, atopic dermatitis, medications, glucocorticoid receptor gene polymorphisms and unpredictable external psychological stressors can affect cortisol concentrations (Kudielka, Hellhammer, &

Wüst, 2009; Lanni et al., 2012). Further, sAA activity is dependent on saliva gland function, parasympathetic and sympathetic influences on protein secretions and saliva flow rate (Cheshire, 2012). In addition to these extraneous factors, children with ASD can move from states of biological under-arousal to over-arousal without a known precipitant (Joosten, Bundy, & Einfeld, 2009; Ozsivadjian, Knott, & Magiati, 2012).

It is possible that reductions in sCort concentrations and sAA activity may have not be associated with SIB reductions. For example, reducing SIB might have acted to lower sCort concentrations and sAA activity. This may have corroborated with the report that SIB may elicit a sense of relief for adults with developmental disabilities (Barrera et al., 2007; Haines, Williams, Brain, & Wilson, 1995a, 1995b; Jones, Congiu, & Stevenson, 1979). Further, based on the paper by Symons et al. (2003), over-arousal may be associated with higher SIB frequencies. As such, it may have been more pertinent to investigate factors that cause over-arousal. Although this was not integrated into the design of the current research, such information may have been gathered from specific questions posed to primary carers during the clinical interview administered in Study 3.

It is also possible in Study 3 that sCort concentrations, sAA activity and SIB frequencies were influenced by the behaviour of other children on the school bus or that listening to music on the school bus radio before and after exposure to the *Sonata Pathetique*. These additional extraneous factors appear to be indicative of conducting research in naturalistic settings.

Despite a report suggesting that the measurement of biomarkers in applied research can result in a greater understanding of how behaviour, physiology and psychosocial factors can impact on behaviour change, the analysis of saliva sampling time-points for the present research was restricted to before and after periods of music listening (White & Mulligan, 2009). This approach was taken because sampling saliva was likely to confound both the naturally occurring SIB and biomarker concentrations/activity.

Future research may consider alternate methods of measuring biological arousal amongst children with LFA. A product known as the *Lifeshirt*, an undergarment worn on the torso of the participant, has been reported to be effective in measuring arousal via electrocardiograph and heart rate from sensors which are placed within the shirt (Wilhelm, Born, Kudielka, Schlotz, & Wüst, 2007). The product is reported to be made of a stretchable fabric, comes in several sizes and is designed to be worn close

to the skin. The *Lifeshirt* has been used in research designed to assess responses of children with LFA to demand conditions (Goodwin et al., 2006). More recently, arousal has been measured via electrodermal activity for sweat response and blood volume pulse (BVP) for beat-to-beat rate derived from sensors attached to the hands of children with ASD by tape and straps to assess responses to demand conditions (Kushki et al., 2013). Further, based on the *Polyvagal Theory* published by Porges (2001), vagal tone (VT) has been reported to be effective in the measurement of ANS activity via heart beat variations which are regulated by the vagal nerve. VT data are collected via electrodes placed on the chest and abdominal skin of child participants to assess the arousal of children with less severe ASD (Levine et al., 2012).

Despite the published efficacy of the above methods, children with LFA may be averse to wearing garments or having electrodes affixed to their hands, chests or abdominal skin via tape or straps due to "hyper- or hyporeactivity to sensory input… adverse response to specific sounds or textures…" features associated with the ASD component of the LFA diagnosis (American Psychiatric Association, 2013, p. 51). For example, participants with severe disabilities such as LFA can be averse to wearing headphones (Hooper et al., 2012).

Such aversions may be minimised by researchers by including a period of sensory desensitisation to the chosen apparatus. Such desensitisation may achieve a stable arousal baseline prior to the participant commencing research. Further, the inclusion of a standardised assessment such as the *Sensory Profile 2* as published by Dunn (2014) may be useful in determining possible sensory aversion factors associated with the application of such apparatus prior to commencing research. However, it must be noted that behavioural and arousal responses of children with LFA can be unpredictable under demand conditions despite the application of desensitisation protocols.

An additional limitation within the present research was the insufficient specificity contained within the inclusion criteria for both Study 2 and Study 3. The inclusion criteria for Study 2 was males with a formal diagnosis of ASD and ID consistent with a diagnosis of LFA. The inclusion criteria for Study 3 was a formal diagnosis of ASD and ID consistent with a diagnosis of LFA in addition to the exhibition of high SIB occurrences. Administering an autism screening tool such as the Autism Diagnostic Observation Schedule (ADOS) or Autism Diagnostic Interview (ADI) may have ensured that each participant met the criteria prior to entry (Bitsika et

al., 2015). Having not accounted for these factors, the heterogeneity of LFA may have confounded results.

It must be noted that, applying more specific inclusion criteria for research including children with LFA can increase the time required to recruit an appropriate number of participants. As was the case in Study 3, after gaining ethical approval via numerous human research ethics committees, five participants were initially deemed suitable. However, only three participants were deemed appropriate for inclusion after further screening revealed that the SIB occurrences of two participants were not frequent enough to be observable via PIR.

As such, SIB frequency assessment scales may have been used in Study 3. The Aberrant Behavior Checklist–Irritability subscale (ABC-I), Questions About Behavioural Function (QABF) scale, and the Childhood Autism Rating Scale (CARS) (Marcus et al., 2011; Paclawskyj et al., 2000; Schopler at al., 1980) may have been incorporated. As previously noted, a comprehensive FA would have accurately identified SIB function.

Further, the diagnostic and behavioural heterogeneity of LFA can influence research results due to the various ASD, ID, and behavioural presentations of people with LFA (Cheung et al., 2010). For example, people with a diagnosis of ASD can meet 5, 6, or 7 criteria with three possible severity levels. Meeting a diagnosis of ID can indicate cognitive deficits ranging from a FSIQ score of 70 points and below on a standardised assessment (American Psychiatric Association, 2013). Regarding Study 3, both Bill and Harry presented with ear-blocking SIB. However, Bill's MAS results indicated that his ear-blocking SIB was motivated by a sensory factor whereas Harry's was motivated by both sensory and tangible factors. The degree of heterogeneity in LFA can threaten assumptions of homogeneity in group research designs such as Study 2 and generalisability in single-case research designs such as Study 3.

Lastly, treatment integrity data may have been collected and reported to reflect the extent to which Study 2 and Study 3 were implemented as planned. This would have improved the internal validity. As such, *adherence* data (to what extent were the specific intervention steps completed), *quality* data (to what standard were the steps of the intervention completed), and *exposure* data (to what extent the participants were exposed to the independent variable) would have been collected and reported for treatment integrity.

7.6 Recommendations for Future Research

Regarding music-based interventions, Fancourt et al. (2014) reported that future studies should focus on understanding the mechanisms of music to determine its maximum potential. As such, employing methods of selecting music for music-based interventions, such as Study 1, may improve the assessment of treatment effect through more accurate replication. Further, applying such methodologies may generalise to music-based interventions for people with conditions with other than LFA who also have an ID component which may also impair their ability to self-select music.

In Study 3, no associations were found between sAA and sCort. However, sAA activity can be more immediately detected than sCort concentrations (Quas, 2011). In research, on average sAA activity peaks approximately 10 minutes and sCort concentration peaks approximately 20 minutes after exposure to a demand condition in research (White & Mulligan, 2009). This time course variance may have influenced the analysis of associations between the two biomarkers. Future research may consider the use of time-series analyses to account for such biological time course variance.

Lastly, including a comprehensive FA in single-case research designs that analyse behavioural and biological data, may increase the likelihood treatment efficacy due to the selection of an appropriate treatment modality. Further, it is recommended that school teachers may be included as FA respondents to improving the accuracy of data.

7.7 Conclusion

This research was designed to assess whether there was an association between music listening, arousal and SIB for boys with LFA. Although an association was identified between music listening and arousal in a controlled environment, no associations were found when applied in a naturalistic setting. This may have been due to numerous uncontrolled variables. Despite this, the present research provided some novel methodological approaches which may be useful for future research, particularly the selection of music for music-based interventions and the use of biomarkers to assess arousal in boys with LFA. It is hoped that the findings from this research will positively contribute to the lives of those with LFA, particularly those who exhibit SIB.

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Appendix A. DSM-III Diagnostic Criteria for Infantile Autism (American Psychiatric Association, 1980, pp. 89-90).

Diagnostic criteria for Infantile Autism

A. Onset before 30 months of age.

B. Pervasive lack of responsiveness to other people (autism).

C. Gross deficits in language development.

D. If speech is present, peculiar speech patterns such as immediate and delayed echolalia, metaphorical language, pronominal reversal.

E. Bizarre responses to various aspects of the environment, e.g. resistance to change, peculiar interest in or attachments to animate or inanimate objects.

F. Absence of delusions, hallucinations, loosening of associations, and incoherence as in Schizophrenia.

299.00 Infantile Autism, Full Syndrome Present

Currently meets the criteria for Infantile Autism

299.01 Infantile Autism, Residual State

Diagnostic criteria for Infantile Autism, Residual State

A. Once had an illness that met the criteria for Infantile Autism.

B. The current clinical picture no longer meets the full criteria for Infantile Autism, but signs of the illness have persisted to the present, such as oddities of communication and social awkwardness.

Appendix B. DSM-III-R Diagnostic Criteria for Autistic Disorder (American Psychiatric Association, 1987, pp. 38-39).

Diagnostic criteria for 299.00 Autistic Disorder

At least eight of the following sixteen items are present, these include at least two items from A, one from B, and one from C.

Note: Consider a criterion to be met *only* if the behavior is abnormal for the person's developmental level.

A. Qualitative impairment in reciprocal social interaction as manifested by the following: (The examples within parentheses are arranged so that those first mentioned are more likely to apply to younger or more handicapped, and the later ones, to older or less handicapped, persons with this disorder.)
   (1) marked lack of awareness of the existence or feelings of others (for example, treats a person as if that person were a piece of furniture; does not notice another person's distress; apparently has no concept of the need of others for privacy
   (2) no or abnormal seeking of comfort at times of distress (e.g., does not come for comfort even when ill, hurt, or tired; seeks comfort in a stereotyped way, for example, says "cheese, cheese, cheese" whenever hurt)
   (3) no or impaired imitation (e.g., does not wave bye-bye; does not copy parent's domestic activities; mechanical imitation of others' actions out of context)
   (4) no or abnormal social play (e.g., does not actively participate in simple games; prefers solitary play activities; involves other children in play only as "mechanical aids")
   (5) gross impairment in ability to make peer friendships (e.g., no interest in making peer friendships; despite interest in making friends, demonstrates lack of understanding of conventions of social interaction, for example, reads phone book to uninterested peer)

B. Qualitative impairment in verbal and nonverbal communication and in imaginative activity, as manifested by the following: (The examples within parentheses are arranged so that those first mentioned are more likely to apply to younger or more handicapped, and the later ones, to older or less handicapped, persons with this disorder.)

(1) no mode of communication, such as: communicative babbling, facial expression, gesture, mime, or spoken language

(2) markedly abnormal nonverbal communication, as in the use of eye-to-eye gaze, facial expression, body posture, or gestures to initiate or modulate social interaction (for example, does not anticipate being held, stiffens when held, does not look at the person or smile when making a social approach, does not greet parents or visitors, has a fixed stare in social situations)

(3) absence of imaginative activity, such as play-acting of adult roles, fantasy character or animals; lack of interest in stories about imaginary events

(4) marked abnormalities in the production of speech, including volume, pitch, stress, rate, rhythm, and intonation (for example, monotonous tone, question-like melody, or high pitch)

(5) marked abnormalities in the form or content of speech, including stereotyped and repetitive use of speech (for example, immediate echolalia or mechanical repetition of a television commercial); use of "you" when "I" is meant (for example, using "You want cookie?" to mean "I want a cookie"); idiosyncratic use of words or phrases (for example, "Go on green riding" to mean "I want to go on the swing"); or frequent irrelevant remarks (for example, starts talking about train schedules during a conversation about sports)

(6) marked impairment in the ability to initiate or sustain a conversation with others, despite adequate speech (for example, indulging in lengthy monologues on one subject regardless of interjections from others)

C. Markedly restricted repertoire of activities and interests as manifested by the following:

(1) stereotyped body movements (for example, hand flicking or twisting, spinning, head-banging, complex whole-body movements)

(2) persistent preoccupation with parts of objects (for example, sniffing or smelling objects, repetitive feeling of texture of materials, spinning wheels of toy cars) or attachment to unusual objects (for example, insists on carrying around a piece of string)

(3) marked distress over changes in trivial aspects of environment for example, when a vase is moved from usual position

(4) unreasonable insistence on following routines in precise detail (for example,

insisting that exactly the same route always be followed when shopping)

(5) markedly restricted range of interests and a preoccupation with one narrow interest, e.g., interested only in lining up objects, in amassing facts about meteorology, or in pretending to be a fantasy character

D. Onset during infancy or early childhood.

Specify if childhood onset (after 36 months of age)

Appendix C. DSM-IV Diagnostic Criteria for Autistic Disorder (American Psychiatric Association, 1994, pp. 70-71).

Diagnostic criteria for 299.00 Autistic Disorder

A. A total of six (or more) items from (1), (2), and (3), with at least two from (1), and one each from (2) and (3):

(1) qualitative impairment in social interaction, as manifested by at least two of the following:

a) marked impairment in the use of multiple nonverbal behaviors such as eye-to-eye gaze, facial expression, body postures, and gestures to regulate social interaction

b) failure to develop peer relationships appropriate to developmental level

c) a lack of spontaneous seeking to share enjoyment, interests, or achievements with other people (e.g., by a lack of showing, bringing, or pointing out objects of interest)

d) lack of social or emotional reciprocity

(2) qualitative impairments in communication as manifested by at least one of the following:

(a) delay in, or total lack of, the development of spoken language (not accompanied by an attempt to compensate through alternative modes of communication such as gesture or mime)

(b) in individuals with adequate speech, marked impairment in the ability to initiate or sustain a conversation with others

(c) stereotyped and repetitive use of language or idiosyncratic language

(d) lack of varied, spontaneous make-believe play or social imitative play appropriate to developmental level

(3) restricted repetitive and stereotyped patterns of behavior, interests, and activities, as manifested by at least one of the following:

(a) encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus

(b) apparently inflexible adherence to specific, non-functional routines or rituals

(c) stereotyped and repetitive motor mannerisms (e.g., hand or finger flapping or

twisting, or complex whole-body movements)

(d) persistent preoccupation with parts of objects

B. Delays or abnormal functioning in at least one of the following areas, with onset prior to age 3 years: (1) social interaction, (2) language as used in social communication, or (3) symbolic or imaginative play.

C. The disturbance is not better accounted for by Rett's Disorder or Childhood Disintegrative Disorder.

Appendix D. DSM-IV-TR Diagnostic Criteria for Autistic Disorder (American Psychiatric Association, 2000, p. 75).

Diagnostic criteria for 299.00 Autistic Disorder

A. A total of six (or more) items from (1), (2), and (3), with at least two from (1), and one each from (2) and (3):

(1) qualitative impairment in social interaction, as manifested by at least two of the following:

(a) marked impairment in the use of multiple nonverbal behaviors such as eye-to-eye gaze, facial expression, body postures, and gestures to regulate social interaction

(b) failure to develop peer relationships appropriate to developmental level

(c) a lack of spontaneous seeking to share enjoyment, interests, or achievements with other people (e.g., by a lack of showing, bringing, or pointing out objects of interest)

(d) lack of social or emotional reciprocity

(2) qualitative impairments in communication as manifested by at least one of the following:

(a) delay in, or total lack of, the development of spoken language (not accompanied by an attempt to compensate through alternative modes of communication such as gesture or mime)

(b) in individuals with adequate speech, marked impairment in the ability to initiate or sustain a conversation with others

(c) stereotyped and repetitive use of language or idiosyncratic language

(d) lack of varied, spontaneous make-believe play or social imitative play appropriate to developmental level

(3) restricted repetitive and stereotyped patterns of behavior, interests, and activities, as manifested by at least one of the following:

(a) encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus

(e) apparently inflexible adherence to specific, non-functional routines or rituals

(f) stereotyped and repetitive motor mannerisms (e.g., hand or finger flapping or twisting, or complex whole-body movements)

(g) persistent preoccupation with parts of objects

B. Delays or abnormal functioning in at least one of the following areas, with onset prior to age 3 years: (1) social interaction, (2) language as used in social communication, or (3) symbolic or imaginative play.

C. The disturbance is not better accounted for by Rett's Disorder or Childhood Disintegrative Disorder.

Appendix E. DSM-5 Diagnostic Criteria for Autism Spectrum Disorder (ASD) (American Psychiatric Association, 2013, pp. 50-51).

Autism Spectrum Disorder 299.00 (F84.0)

A. Persistent deficits in social communication and social interaction across multiple contexts, as manifested by the following, currently or by history (examples are illustrative, not exhaustive; see text):

   (a) Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions.

   (b) Deficits in nonverbal communicative behaviors used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.

   (c) Deficits in developing, maintaining, and understanding relationships, ranging, for example, from difficulties adjusting behavior to suit various social contexts; to difficulties in sharing imaginative play or in making friends; to absence of interest in peers.

   Specify current severity:

Severity is based on social communication impairments and restricted, repetitive patterns of behavior (see Table).

B. Restricted, repetitive patterns of behavior, interests, or activities, as manifested by at least two of the following, currently or by history (examples are illustrative, not exhaustive; see text):

(a) Stereotyped or repetitive motor movements, use of objects, or speech (e.g., simple motor stereotypies, lining up toys or flipping objects, echolalia, idiosyncratic phrases).

(b) Insistence on sameness, inflexible adherence to routines, or ritualized patterns of verbal or nonverbal behavior (e.g., extreme distress at small changes, difficulties with transitions, rigid thinking patterns, greeting rituals, need to take the same route or eat same food every day).

(c) Highly restricted, fixated interests that are abnormal in intensity or focus (e.g., strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interests).

(d) Hyper- or hyporeactivity to sensory input or unusual interest in sensory aspects of the environment (e.g., apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement).

Specify current severity:

Severity is based on social communication impairments and restricted, repetitive patterns of behavior (see Table).

C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life).

D. Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning.

E. These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual

disability, social communication should be below that expected for general developmental level.

Note: Individuals with a well-established DSM-IV diagnosis of autistic disorder, Asperger's disorder, or pervasive developmental disorder not otherwise specified should be given the diagnosis of autism spectrum disorder. Individuals who have marked deficits in social communication, but whose symptoms do not otherwise meet criteria for autism spectrum disorder, should be evaluated for social (pragmatic) communication disorder.

Specify if:

- **With or without accompanying intellectual impairment**
- **With or without accompanying language impairment**
- **Associated with a known medical or genetic condition or environmental factor** (**Coding note:** Use additional code to identify the associated medical or genetic condition.)
- **Associated with another neurodevelopmental, mental, or behavioral disorder** (**Coding note:** Use additional code[s] to identify the associated neurodevelopmental, mental, or behavioral disorder[s].) **With catatonia** (refer to the criteria for catatonia associated with another mental disorder, pp. 119–120, for definition) (**Coding note:** Use additional code 293.89 [F06.1] catatonia associated with autism spectrum disorder to indicate the presence of the comorbid catatonia.)

Table 2. Severity levels for Autism Spectrum Disorder.

Severity level	Social communication	Restricted, repetitive behaviors
Level 3 "Requiring very substantial support"	Severe deficits in verbal and nonverbal social communication skills cause severe impairments in functioning, very limited initiation of social interactions, and minimal response to social overtures from others. For example, a person with few words of intelligible speech who rarely initiates interaction and, when he or she does, makes unusual approaches to meet needs only and responds to only very direct social approaches.	Inflexibility of behavior, extreme difficulty coping with change, or other restricted/repetitive behaviors markedly interfere with functioning in all spheres. Great distress/difficulty changing focus or action.
Level 2 "Requiring substantial support"	Marked deficits in verbal and nonverbal social communication skills; social impairments apparent even with supports in place; limited initiation of social interactions; and reduced or abnormal responses to social overtures from others. For example, a person who speaks simple sentences, whose interaction is limited to narrow special interests, and who has markedly odd nonverbal communication.	Inflexibility of behavior, difficulty coping with change, or other restricted/repetitive behaviors appear frequently enough to be obvious to the casual observer and interfere with functioning in a variety of contexts. Distress and/or difficulty changing focus or action.
Level 1 "Requiring support"	Without supports in place, deficits in social communication cause noticeable impairments. Difficulty initiating social interactions, and clear examples of atypical or unsuccessful responses to social overtures of others. May appear to have decreased interest in social interactions. For example, a person who is able to speak in full sentences and engages in communication but whose to-and-fro conversation with others fails, and whose attempts to make friends are odd and typically unsuccessful.	Inflexibility of behavior causes significant interference with functioning in one or more contexts. Difficulty switching between activities. Problems of organization and planning hamper independence.

Appendix F. DSM-I Diagnostic Criteria for Mental Deficiency (American Psychiatric Association, 1952, p. 86).

Mental Deficiencies (Codes: 60-62)

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60. MENTAL DEFICIENCIES (FAMILIAL OR HEREDITARY)

- 60.0 Mild
- 60.1 Moderate
- 60.2 Severe
- 60.3 Severity not specified

61. MENTAL DEFICIENCY, IDIOPATHIC

- 61.0 Mild
- 61.1 Moderate
- 61.2 Severe
- 61.3 Severity not specified

62. MENTAL DEFICIENCY (x4)

- 62.0 Severe
- 62.1 Moderate
- 62.2 Mild
- 62.3 Severity not specified

***** If Mongolism is specified, code 325.4

Appendix G. DSM-II Diagnostic Criteria for Mental Retardation (American Psychiatric Association, 1968, pp. 14-22).

Section 3: I: Mental Retardation (Codes: 310-315)

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310. Borderline mental retardation – IQ 68 – 85

311. Mild mental retardation – IQ 52 – 67

312. Moderate mental retardation – IQ 36 – 51

313. Severe mental retardation – IQ 20 – 35

314. Profound mental retardation – IQ under 20

315. Unspecified mental retardation

Clinical Subcategories of Mental Retardation

.0 Following infection and intoxication

.1 Following trauma or physical agent

.2 With disorders of metabolism, growth or nutrition

.3 Associated with gross brain disease (postnatal)

.4 Associated with diseases and conditions due to unknown prenatal influence

.5 With chromosomal abnormality

.6 Associated with prematurity

.7 Following major psychiatric disorder

.8 With psycho-social (environmental) deprivation

.9 With other [and unspecified] condition.

Appendix H. DSM-III Diagnostic Criteria for Mental Retardation (American Psychiatric Association, 1980, pp. 40-41).

Diagnostic criteria for Mental Retardation

A. Significantly sub average general intellectual functioning: an IQ of 70 or below on an individually administered IQ test (for infants, since available intelligence tests do not yield numerical values, a clinical judgement of significant sub average intellectual functioning).
B. Concurrent deficits or impairments in adaptive behaviour, taking the person's age into consideration.
C. Onset before the age of 18.

(If there are behavioral symptoms requiring attention or treatment [e.g., aggressive behavior, self-mutilation, anxiety symptoms] that are not part of another disorder, the non-ICD-9-CM code "1" may be recorded in the fifth digit. Otherwise, code "0".)

Appendix I. DSM-III-R Diagnostic Criteria for Mental Retardation (American Psychiatric Association, 1987, pp. 31-32).

Diagnostic criteria for Mental Retardation

A. Significantly sub average general intellectual functioning: an IQ of 70 or below on an individually administered IQ test (for infants, a clinical judgement of significant sub average intellectual functioning, since available intelligence tests do not yield numerical values).

B. Concurrent deficits or impairments in adaptive functioning, i.e. the person's effectiveness in meeting the standards expected for his or her age by his or her cultural group in areas such as social skills and responsibility, communication, daily living skills, personal independence, and self-sufficiency.

C. Onset before the age of 18.

(If there are behavioral symptoms requiring attention or treatment [e.g., aggressive behavior, self-mutilation, anxiety symptoms] that are not part of another disorder, the non-ICD-9-CM code "1" may be recorded in the fifth digit. Otherwise, code "0".)

Appendix J. DSM-IV Diagnostic Criteria for Mental Retardation (American Psychiatric Association, 1994, p. 46).

Diagnostic criteria for Mental Retardation

A. Significantly sub average general intellectual functioning: an IQ of 70 or below on an individually administered IQ test (for infants, a clinical judgement of significant sub average intellectual functioning).

B. Concurrent deficits or impairments in present adaptive functioning (i.e. the person's effectiveness in meeting the standards expected for his or her age by his or her cultural group) in at least two of the following areas: communication, self-care, home living, social/interpersonal skills, use of community resources, self-direction, functional academic skills, work, leisure, health, and safety.

C. The onset is before age 18 years.

Appendix K. DSM-IV-TR Diagnostic Criteria for Mental Retardation (American Psychiatric Association, 2000, p. 49).

Diagnostic criteria for Mental Retardation

A. Significantly sub average general intellectual functioning: an IQ of 70 or below on an individually administered IQ test (for infants, a clinical judgement of significant sub average intellectual functioning).

B. Concurrent deficits or impairments in present adaptive functioning (i.e. the person's effectiveness in meeting the standards expected for his or her age by his or her cultural group) in at least two of the following areas: communication, self-care, home living, social/interpersonal skills, use of community resources, self-direction, functional academic skills, work, leisure, health, and safety.

C. The onset is before age 18 years.

Appendix L. DSM-5 Diagnostic Criteria for Intellectual Disability (Intellectual Developmental Disorder) (American Psychiatric Association, 2013, p. 33).

Intellectual Disability (Intellectual Developmental Disorder) 319.00

A. Intellectual disability (intellectual developmental disorder) is a disorder with onset during the developmental period that includes both intellectual and adaptive functioning deficits in conceptual, social, and practical domains. The following three criteria must be met:

B. Deficits in intellectual functions, such as reasoning, problem solving, planning, abstract thinking, judgment, academic learning, and learning from experience, confirmed by both clinical assessment and individualized, standardized intelligence testing.

C. Deficits in adaptive functioning that result in failure to meet developmental and sociocultural standards for personal independence and social responsibility. Without ongoing support, the adaptive deficits limit functioning in one or more activities of daily life, such as communication, social participation, and independent living, across multiple environments, such as home, school, work, and community.

D. Onset of intellectual and adaptive deficits during the developmental period.

Appendix M. Study 1: Recruitment Email and Information Letter.

Curtin University
School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

To current or past primary carers of males with autism and an intellectual disability (defined as Low Functioning Autism or LFA),

We are pleased and excited to collaborate with the world renowned concert pianist Mr David Helfgott on an approved study that investigates the potential of listening calming music to reduce arousal and challenging behaviours for males with LFA! If you are, or have ever been the primary carer for a male with LFA you are invited to participate in this online survey! David Helfgott has used his exceptional talents to select and provide the first 2 minutes of 6 solo piano performances for you to listen, assess then rate ... requiring no more than 15 minutes of your time!
Your musical ratings (along with those from other primary carers) will be statistically analysed to identify one musical segment that will be applied in 2 further studies. As you may imagine, this research has the potential to benefit children with Autism, families, schools and therapists.

Participation is as easy as ***clicking the links*** at **Steps 1,2,3**:
Step 1: check that your computer is running the latest version of Adobe Flash Player! For the latest free version ***click the link*** http://get.adobe.com/flashplayer/
Step 2: ***click the link*** http://davidhelfgott.com/news/ opening the story
"David provides piano performances for LFA study" | February 06 2012
Step 3: ***click the link*** https://curtin.asia.qualtrics.com/SE/?SID=SV_295lfBwPfPxiJo0 and complete the survey!

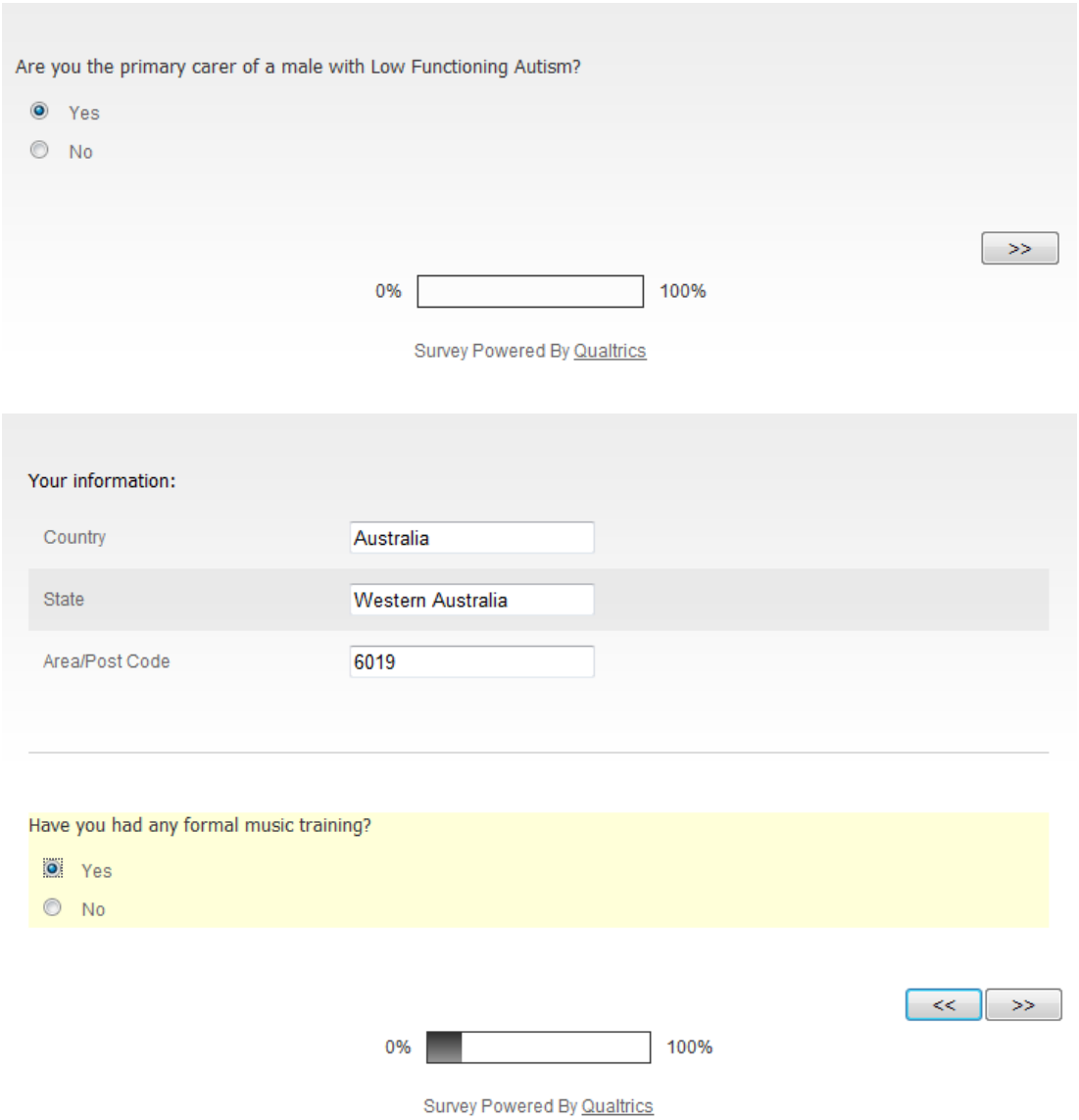
Please forward this email to anyone who may be interested in participating in this survey and/or the following 2 studies:
Study 2 (ACTRN12611001063909) – 30 participants required between July to December, 2012!
In this Randomised Control Trial (RCT), males aged between 6 and 11 years accompanied by their primary carer(s) will attend Curtin University in Perth Western Australia and be randomly assigned to one of 2 groups.
The **no music group** will watch footage of a school bus ride on a big screen television. While watching the footage (14 minutes), self-injury attempts which usually occur for the participants with LFA when on the school bus will be recorded (I say 'attempts' because the participants are required to wear their usual personal protective equipment at all times to ensure no harm). When the footage stops, a saliva sample will be collected from the participant's mouth and analysed for biological indicators of stress such as Cortisol and Alpha Amylase.
The **music group** will undergo the same procedure ... however, the school bus ride footage will include the music chosen from this study (Study 1)! It is hypothesised that the music group will record fewer self-injury attempts and lower levels of stress in saliva than the no music group.

Study 3 (ACTRN12611001064998) – 6 participants required between January to June 2013!
Also an RCT, Study 3 replicates Study 2 ... with one major difference ... it happens on a **real school bus**.
For any questions or if you live in Perth, Western Australia and would like to register your interest for Study 2 or Study 3 , please contact Jeremy Marriott on (M) 0412 320 762 or Email: jeremy.k.marriott@postgrad.curtin.edu.au
Thank you for your time, consideration and contribution.
Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

If you require further information please contact Prof Jan Piek (08) 9266 7990 at the School of Psychology and Speech Pathology, Curtin University. If you wish to contact someone independent of the study with any questions or concerns, please contact the Office or Research and Development at Curtin University on (08) 9266 7863.This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number **HR138/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning (08) 9266 2784 or by emailing hrec@curtin.edu.au

Appendix N. Study 1: Qualtrics Survey Software – Survey



Please describe your formal music training (total years, type of training ... etc.):

10 years

Information and Consent Form
(Please read this important information)

An introduction ...
Study 1 investigates the potential of Receptive Music Therapy (music listening) to reduce challenging behaviours for males with Low Functioning Autism (LFA) while traveling to school by bus! As a primary carer for a male with LFA, we invite you to choose the music in the first of 3 studies! Mr David Helfgott has selected and provided 6 solo piano performances for you to listen, assess then rate. Your music ratings (along with those from other primary carers) will be statistically analysed to identify one piece that will be used in 2 further studies. As you can imagine, this research has the potential to benefit children with Autism, their families and schools.

Things to know before you start ...
1) We recommend you install the latest free version of Adobe Flash Player from http://get.adobe.com/flashplayer/ before you start. This will enable you to access the Music Player (versions pictured below).
2) This is an untimed survey, so please take your time and enjoy the music.
3) << >> will appear at the bottom of each survey screen. So, feel free to travel forwards or backwards as much as you like.
4) Please listen to the whole 2 minutes of each musical piece to get the full experience.
5) Each of the 6 musical pieces are presented to you in a specific order. Please rate them in this order.
6) Please play only one piece of music at a time ... listening to more than one at a time can be rather confusing!
7) The music is in MP3 format. This is reported to be the most widely used and easily played audio format. If you have trouble playing the music from you computer, please contact jeremy.k.marriott@postgrad.curtin.edu.au for assistance.*
8) Please make sure that your computer sound system is turned on, volume is up and speakers are in good working order before playing the music ... headphones may also be helpful!

* There may be a minor delay after clicking play ... usually the time it takes for your computer to download the music. Please be patient ... it will be worth the wait!

The Music Player ...

To play the music selected and provided by David Helfgott, you will need to operate a Music Player (one for each piece). Examples of some different versions of music players (dependent on which browser you prefer) include:

a) Safari browser (Mac, iPhone, iPad):

b) Firefox browser (Mac):

c) Internet Explorer browser (PC)

Which ever internet browser you use, clicking the '**Play**' button will start the music.

Your participation!
In the order they are presented (working from the top to the bottom of the page ... you will need to scroll down to rate all 6 performances), please listen then rate each performance on the scale from "***Not at all Calming***", "***Moderately Calming***" to "***Very Calming***" based on how calming each piece would be if the male you care for with LFA listened. David graciously selected the first 2 minutes of each performance to minimise the total time needed to complete the survey. It should take no more than 15 minutes in total!

At the end of the survey, only if David gives you the "**Thumbs Up**", click the **right** of these **arrow** << >> to record your response!

And then ...
As a result of this study, listening to calming music may emerge as a non-invasive therapy option for reducing or even avoiding challenging behaviour for people with LFA.
The results of Study 1 and 2 further studies will appear in scientific journals, at Curtin University and in collaboration with David Helfgott. Regardless of where the results appear, we would like you to know that your identity, information and music ratings are and will remain anonymous. As a matter of fact, the only people with access to your information are the research team. Your responses will be password protected and stored securely by Qualtrics and the research team, statistically analysed then deleted.

Your involvement and any questions ...
We would like you to know that your involvement is completely voluntary. We value your right to withdraw at any stage without penalty. If you would like to withdraw, please simply close your Internet browser. If you would like to provide feedback, your confidentiality will be maintained, please contact jeremy.k.marriott@postgrad.curtin.edu.au

Study 2 and Study 3 ... any participants?
The most calming music chosen from this study (Study 1) will be used in Study 2 and Study 3. Both have achieved registration with the Australian New Zealand Clinical Trials Registry (ANZCTR). We wanted to tell you about these studies because:
1) We think it is important for you to know how the music you rate will be used.
2) We are currently recruiting for participants for Study 2 and Study 3!

Study 2 (ACTRN12611001063909) - 30 participants required between July to December, 2012!
In this Randomised Control Trial (RCT), males aged between 6 and 11 years accompanied by their primary carer(s) will attend Curtin University in Perth Western Australia and be randomly assigned to one of 2 groups.
The **non-music group** will watch footage of a school bus ride on a big screen television. While watching the footage (13 minutes) the frequency of self-injury attempts will be recorded (I say 'attempts' because the participant will wear personal protective equipment to ensure no harm). When the footage stops, a saliva sample will be collected from the participants mouth and analysed for biological indicators of stress such as Cortisol, Alpha Amylase and Chromgranin A.
The **music group** will undergo the same procedure ... however, the school bus ride footage will include the music chosen from this study (Study 1)! It is hypothesised that the music group will record a lower frequency of self-injury attempts and lower levels of stress in saliva than the non-music group.

Study 3 (ACTRN12611001064998) - 6 participants required between January to June 2013!
Also an RCT, Study 3 replicates Study 2 ... with one major difference ... it happens on a **real school bus**.

If you live in Perth, Western Australia and would like to register your interest for Study 2 or Study 3, please contact Jeremy Marriott on (M): 0412 320 762 or Email: jeremy.k.marriott@postgrad.curtin.edu.au

Anything else ...
If you require further information please contact Prof Jan Piek (08) 9266 7990 at the School of Psychology and Speech Pathology, Curtin University. If you wish to contact someone independent of the study with any questions or concerns please contact the Office or Research and Development at Curtin University on (08) 9266 7863.
This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number **HR138/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning (08) 9266 2784 or by emailing hrec@curtin.edu.au

---

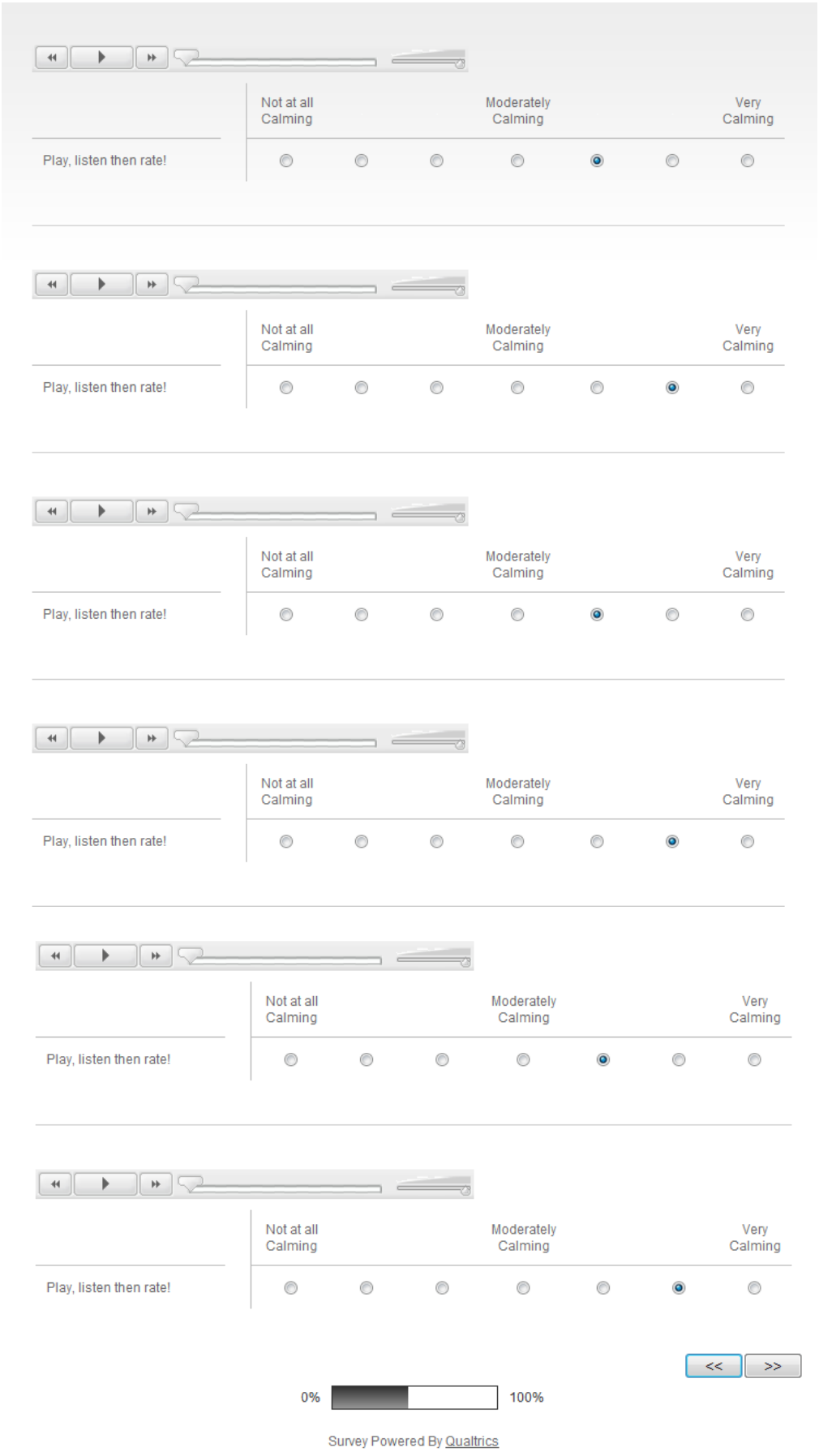
I have read and understood the above Information and Consent form and have decided of my own free will to participate in this study (Study 1).

- (•) Yes
- ( ) No

<< >>

0% 100%

Survey Powered By Qualtrics


Not at all Calming
Moderately Calming
Very Calming
Play, listen then rate!
Not at all Calming
Moderately Calming
Very Calming
Play, listen then rate!
Not at all Calming
Moderately Calming
Very Calming
Play, listen then rate!
Not at all Calming
Moderately Calming
Very Calming
Play, listen then rate!
Not at all Calming
Moderately Calming
Very Calming
Play, listen then rate!
<<
>>
0%
100%
Survey Powered By Qualtrics

Thumbs Up!

Thank you for participating in Study 1 of Music, Arousal and Self-Injurious Behaviour: A 3-Stage Mediating Model for Children with Low Functioning Autism by Jeremy Marriott of Curtin University, Perth, Western Australia.Your time and participation is most appreciated not only by the research team, but by those with Autism, their families and schools.

Please click the **right arrow** to register your response and tour
http://www.davidhelfgott.com

<< >>

0% 100%

Survey Powered By Qualtrics

Appendix O. Study 2: Recruitment Flyer.

Can *Music* calm children with Autism?

Jeremy Marriott - PhD candidate | Autism practitioner

&

David Helfgott - Pianist | subject of the 1996 movie *Shine*...

... are recruiting NOW! Contact Jeremy to find out more!

☎ 0412 320 762

jeremy.k.marriott@postgrad.curtin.edu.au

Curtin University

HREC | HR138/2011 Study 2 | ACTRN12611001063909 Study 3 | ACTRN12611001064998

Curtin University is a trademark of Curtin University of Technology. CRICOS Provider Code 00301J (WA), 02637B (NSW)

If you require further information please contact Prof Jan Piek (08) 9266 7690 at the School of Psychology and Speech Pathology, Curtin University. you wish to contact someone independent of the study with any questions or concerns please contact the Office or Research and Development at Curtin University on (08) 9266 7863.This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR138/2011). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning (08) 9266 2784 or by emailing hrec@curtin.edu.au

Appendix P. Study 2: Parent Information Letter and Consent Form.

10 October 2013

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

Dear Parent(s),

The School of Psychology and Speech Pathology and the University Human Research Ethics Committee at Curtin University along with the Department of Education and the Public Transport Authority WA – School Bus Services have approved 2 research projects which investigate the potential for calming music to reduce arousal (**Study 1**) and Self-Injurious Behaviour (SIB) (**Study 2**) for boys with autism and an intellectual disability (defined as Low Functioning Autism or LFA). This doctoral research is entitled *Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism.* Funded by Curtin University and the Department of Health WA, I am currently asking for school-aged males to participate.

Study 2 (registered with the Australian New Zealand Clinical Trials Registry ACTRN12611001063909) 30 school-aged males with LFA are invited to attend Curtin University Bentley Campus on one occasion to watch a 14-minute video clip of a school bus ride with or without calming piano music. When the video and music stops you will be asked to collect small saliva samples from your son's mouth (I will be there to help if needed). I will then analyse the saliva in a lab for 3 biological markers of stress/arousal.

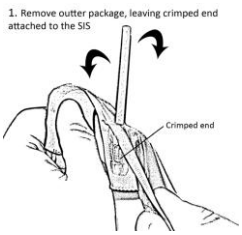
I invite you to register your interest.

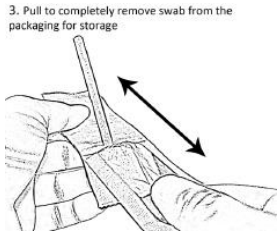
Study 3 (ACTRN12611001064998) 6 school-aged males with LFA who exhibit self-injurious behaviours are invited to listen to calming piano music for the last 14 minutes of their usual morning school bus ride. Saliva will be collected and analysed as per Study 1 and footage will be recorded and analysed for self-injurious behaviour attempt frequencies. The word *attempts* are used to highlight that your son will be required to wear any personal protective equipment usually worn on the bus at all times. School staff will collect the saliva samples.

I invite you to register your interest.

Just so you know what is involved in the saliva collection, please find a protocol below (Salimetrics, 2011, p. 8-9):

1. Peel open the outer package and remove the device.
2. Securely hold one end of the device … place the other end under the child's tongue ... it may only be possible to collect pooling saliva (often at the corners of the mouth). Re-introduce into the mouth as needed until the lower third of the swab is saturated (60-90 seconds total).
3. Remove the swab for storage.





Your participation, although voluntary, will play a major part in the assessment of this innovative, safe and non-invasive experimental treatment. Please know that you and your son's information will be de-identified, confidential and we support your withdrawal at any time without prejudice or penalty. If you like, I am happy to provide a summary report of the research findings when it is finished. To *register your interest*, please complete the **Consent Form** for either **Study 1** or **Study 2** and return fax it (or sign, scan and email). I can then contact you to answer any questions and arrange the best time to meet. I welcome your contact at any stage.

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

If you require further information please contact Prof Jan Piek (08) 9266 7990 at the School of Psychology and Speech Pathology, Curtin University. If you wish to contact someone independent of the study with any questions or concerns, please contact the Office or Research and Development at Curtin University on (08) 9266 7863.This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number **HR138/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning (08) 9266 2784 or by emailing hrec@curtin.edu.au

Curtin University
School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

I/we _____ _____ agree to participate in

Study 2 of the experimental research entitled:

Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism conducted by Jeremy Marriott of Curtin University.

I/we give written consent that:

- I/we have received the information letter and understand the purpose and procedures of Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism.
- I/we have been given the opportunity to ask and have answered questions prior to consenting.
- I/we confirm that my/our son has Autism and an intellectual disability.
- I/we understand that Jeremy Marriott has a National Police Certificate and Working with Children Card.
- I/we understand that involvement is confidential between the experimenter and will not be revealed without written consent.
- I/we understand the importance of my/our son not deviating from regular treatment for the duration of the study.
- I/we am/are aware that I/we will be required to:
    a. Participate in a structured telephone interview.
    b. Allow the researcher access to my/our home on one occasion and the school bus on 4 occasions to support the collection of 5 saliva samples per occasion from my/our son's mouth using a special swab.
    c. Allow the researcher to collect and analyse saliva samples for this research and for potential use in future studies with the researcher permission. I/we understand that my/our son's data will be de-identified.
    d. I/we am/are aware that my son will be identified with a 3-digit code to maintain anonymity.
- I/we am/are aware that all data will be treated with respect, privacy and will be held securely by the schools of Psychology and Biomedical Sciences at Curtin University for at least 5 years then destroyed.
- I/we acknowledge that information generated from this research will retain my/our and my/our son's anonymity and be distributed within Curtin University, academic journals and other venues.
- I/we give consent for the researcher to record information and understand that participation is voluntarily and can be withdrawn at any time without prejudice/penalty.
- I/we acknowledge that I/we can be provided a copy this form, the final report or any other information regarding this study upon request.
- I/we acknowledge that the potential benefits of this research are in excess of the risks of participation.

☐ **YES**, _____ I/we **would like to** register my/our and my/our son's participation for **Study 2** of Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism from 2012-2013.

Child Name: _____

Parent/Primary Carers Name(s): _____

Parent/Primary Carers Signature(s): _____

Contact Phone Number:

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

To return this form please:
Scan & email: jeremy.k.marriott@postgrad.curtin.edu.au
or
Fax: (08) 9266 3178
Thank you for your time.

If you require further information please contact Prof Jan Piek (08) 9266 7990 at the School of Psychology and Speech Pathology, Curtin University. If you wish to contact someone independent of the study with any questions or concerns, please contact the Office or Research and Development at Curtin University on (08) 9266 7863.This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number **HR138/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning (08) 9266 2784 or by emailing hrec@curtin.edu.au

Appendix Q. Study 2: Parent Information Letter and Social Story.

Curtin University

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism

Appointment: Study 2 (ACTRN12611001063909)
Location: Building 500, Curtin University – Bentley Campus (just off Manning road)
Day:
Time: 8:15am
Phone: 0412 320 762

Important instructions:

1) PARTICIPANT is required to not have any breakfast till the trial is complete.
2) I have an arrangement with the Aroma Cafe on site to provide some coffee/juice and muffin for breakfast.
3) PARTICIPANT is required to not brush or floss his teeth on the morning of the trial.
4) Please know that PARTICIPANT will be required to stay at Curtin University under your (and my) supervision for at least 20 minutes after the trial has finished. So, please feel free to bring something that he may like to do during this time. Please feel free to bring anything PARTICIPANT likes to do during these 20 minutes, either in the research room, foyer or outside ... I can even play a movie on the big screen!! ... Whatever you like.

I will assist you to collect saliva samples using a special child swab:

a) Immediately before the school bus ride footage
b) Immediately after the school bus ride footage
c) 5 minutes after the school bus ride footage
d) 10 minutes after the school bus ride footage
e) 20 minutes after the school bus ride footage

Just so you know what is involved in the collection of saliva:

4. Gloves are available if you like.
5. Securely hold one end of the swab and try to put it under your son's tongue ... it may only be possible to collect pooling saliva (often at the corners of the mouth).
6. Give the sample to Jeremy.

The location...

The building...

The car park...

Your own parking space ...

The school bus simulator...

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

Appendix R. Study 3: Ethics Approvals, Information Letters and Consent Forms.

Study 3: Curtin University Ethics Clearance

Curtin University

Memorandum

To	Professor Jan Piek, School of Psychology and Speech Pathology
From	A/Professor Stephan Millett, Chair, Human Research Ethics Committee
Subject	Protocol Approval **HR 138/2011**
Date	25 October 2011
Copy	Mr Jeremy Kent Marriott School of Psychology and Speech Pathology Dr Robert Kane School of Psychology and Speech Pathology Dr Cyril Mamotte School of Psychology and Speech Pathology

Office of Research and Development

Human Research Ethics Committee

TELEPHONE 9266 2784
FACSIMILE 9266 3793
EMAIL hrec@curtin.edu.au

Thank you for providing the additional information for the project titled "*Music, arousal and self-injurious behaviour: a 3-Stage Mediating Model for children with Low Functioning Autism*". The information you have provided has satisfactorily addressed the queries raised by the Committee. Your application is now **approved**.

- You have ethics clearance to undertake the research as stated in your proposal.
- The approval number for your project is **HR 138/2011.** *Please quote this number in any future correspondence.*
- Approval of this project is for a period of twelve months **24-10-2011** to **24-10-2012**. To renew this approval a completed Form B (attached) must be submitted before the expiry date **24-10-2012**.
- If you are a Higher Degree by Research student, data collection must not begin before your Application for Candidacy is approved by your Faculty Graduate Studies Committee.
- The following standard statement **must be** included in the information sheet to participants:
  This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number HR 138/2011). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning 9266 2784 or by emailing hrec@curtin.edu.au.

Applicants should note the following:

It is the policy of the HREC to conduct random audits on a percentage of approved projects. These audits may be conducted at any time after the project starts. In cases where the HREC considers that there may be a risk of adverse events, or where participants may be especially vulnerable, the HREC may request the chief investigator to provide an outcomes report, including information on follow-up of participants.

The attached **FORM B** should be completed and returned to the Secretary, HREC, C/- Office of Research & Development:
When the project has finished, or

- If at any time during the twelve months changes/amendments occur, or
- If a serious or unexpected adverse event occurs, or
- 14 days prior to the expiry date if renewal is required.
- An application for renewal may be made with a Form B three years running, after which a new application form (Form A), providing comprehensive details, must be submitted.

Regards,

A/Professor Stephan Millett
Chair Human Research Ethics Committee

Study 3: Department of Education Ethics Clearance

Government of **Western Australia**
Department of **Education**

Your ref :
Our ref : D12/0270293
Enquiries :

Mr Jeremy K. Marriott
School of Psychology and Disability
Curtin University
GPO Box U1987
PERTH WA 6845

Dear Jeremy

Thank you for your completed application received 14 February 2012 to conduct research on Department of Education sites.

The focus and outcomes of your research project, *Music, Arousal and Self-Injurious Behaviour: A 3 Stage Mediating Model for Children with Low Functioning Autism,* are of interest to the Department. I give permission for you to approach site managers to invite their participation in the project as outlined in your application. It is a condition of approval, however, that upon conclusion the results of this study are forwarded to the Department at the email address below.

Consistent with Department policy, participation in your research project will be the decision of the schools invited to participate, individual staff members, the children in those schools and their parents. A copy of this letter must be provided to site managers when requesting their participation in the research. Researchers are required to sign a confidential declaration and provide a current Working with Children Check upon arrival at the Department of Education site.

Responsibility for quality control of ethics and methodology of the proposed research resides with the institution supervising the research. The Department notes a copy of a letter confirming that you have received ethical approval of your research protocol from the Curtin University Human Research Ethics Committee.

Any proposed changes to the research project will need to be submitted for Department approval prior to implementation.

Please contact Ms Allison McLaren, A/Evaluation Officer, on (08) 9264 5512 or researchandpolicy@det.wa.edu.au if you have further enquiries.

Very best wishes for the successful completion of your project.

Yours sincerely

ALAN DODSON
DIRECTOR
EVALUATION AND ACCOUNTABILITY

4 May 2012

151 Royal Street, East Perth Western Australia 6004

Study 3: PTA-SBS Ethics Clearance

Monday, 18 June 2012 3:19

Jeremy,

Thank you for clarifying the scope of your work. On the basis of this clarification I am happy to approve your access to 'orange' school buses for the purposes of conducting your research project. As these services are contracted, you will need the bus contractor's agreement to participate as well since PTA does not own the buses and are not contractually obliged to require them to participate.

In order to facilitate your access to our contracted 'orange' school buses, can you please provide me with a list of 'orange' school bus services you wish to access based on the children you already have for your research project. PTA will email the contractors concerned to inform them of your research project.

On that note can I ask that you change your letter and address it to 'school bus contractor' in lieu of bus driver as the contractor owns the bus and the driver is normally an employee of the contractor. Can you also remove the address block as I intend to email your letter to the contractor on your behalf. The email will also direct contractor to liaise directly with you to give you their consent to be part of the trial. If for some reason you are not contacted please let me know.

For administrative purposes can you provide me with a list of all personnel who will be travelling on the buses for filming and a scanned copy of their Working with Children Notice. This must be done before anyone can get onboard a bus.

If you require any assistance during your research project can you please liaise with Karen Ferrier 9326 2607 as necessary.

Regards

John Bailly | Manager
School Bus Services
Public Transport Authority WA | PO Box 8125, Perth Business Centre, WA, 6000
Phone: 9326 2654 | **Fax: 9326 2862** | eMail : John.Bailly@pta.wa.gov.au

Appendix S. Study 3: Information Letters and Consent Forms.

Curtin University

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

Dear PRIMARY CARER,

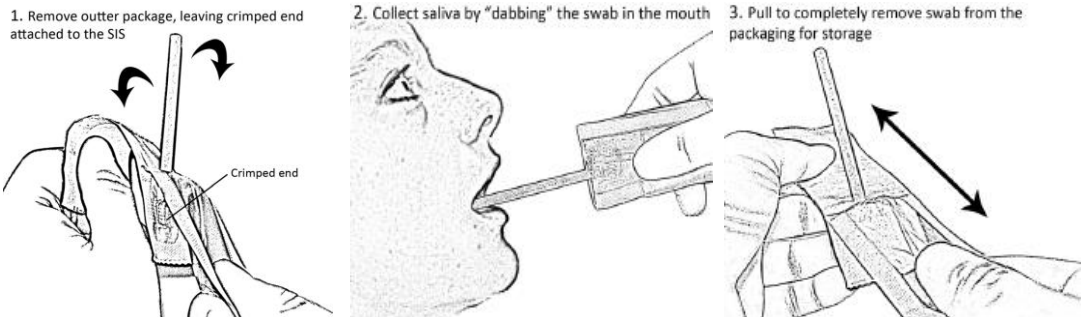
The School of Psychology and Speech Pathology and the University Human Research Ethics Committee at Curtin University along with the Department of Education and the Public Transport Authority WA – School Bus Services have approved 2 research projects which investigate the potential for calming music to reduce arousal and Self-Injurious Behaviour (SIB) for boys with autism and an intellectual disability (defined as Low Functioning Autism or LFA). This doctoral research is entitled *Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism* (ACTRN12611001064998). Enabled by Curtin University and the Department of Health WA funding, I am currently offering participation to school-aged males.

Study 3: Up to 6 school-aged males with Low Functioning Autism who exhibit self-injurious behaviours are invited to listen to calming piano music for the last 14 minutes of their usual journey to school. Saliva samples will be collected before, immediately after, 5, 10 and 20 minutes after arriving at school and analysed for 3 biomarkers of stress. In addition, footage will be recorded (exclusively of the participant and for research purposes only) and analysed for self-injurious behaviour attempts both on the bus and for the first 20 minutes of their time at school. The word *attempt* is used to highlight that your son will be required to wear any personal protective equipment usually worn on the bus at all times.

I invite you to register your interest.

Just so you know what is involved in the saliva collection, please find a protocol below (Salimetrics, 2011, p. 8-9):

1. Peel open the outer package and remove the device.
2. Securely hold one end of the device … place the other end under the child's tongue ... it may only be possible to collect pooling saliva (often at the corners of the mouth). Re-introduce into the mouth as needed until the lower third of the swab is saturated (60-90 seconds total).
3. Remove the swab for storage.



You and your son's participation, although voluntary, will play a major part in the assessment of this innovative, safe and non-invasive experimental treatment. Please know that all information will be de-identified, confidential and I support your withdrawal at any time without prejudice or penalty. If you like, I am happy to provide a summary report of the research findings when it is finished. To *register your interest*, please complete the **Consent Form** and return fax it (or sign, scan and email). I can then contact you to answer any questions before making further arrangements. I welcome your contact at any stage.

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

Study 3: Primary Carer Consent Form.

Curtin University

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

I/we **PRIMARY CARER NAME** agree to participate in **Study 3** of the experimental research entitled: **Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism** conducted by Jeremy Marriott of Curtin University.
I/we give written consent that:

- I/we have received the information letter and understand the purpose and procedures of Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism.
- I/we have been given the opportunity to ask and have answered questions prior to consenting.
- I/we confirm that my/our son has Autism and an intellectual disability.
- I/we understand that the researcher has a current National Police Certificate and Working with Children Card.
- I/we understand that involvement is confidential and identities will not be revealed without written consent.
- I/we understand the importance of my/our son not deviating from regular treatment for the duration of the study.
- I/we am/are aware that I/we will be required to:
    a. Participate in a structured telephone/in-person interview.
    b. Allow the researcher access to the school bus/vehicle on 4 occasions to collect 5 saliva samples and for one day each week over 4 consecutive weeks from my/our son's mouth using a special swab.
    c. Allow the researcher to record footage exclusively of my/our son both on the school bus and for the first 20 minutes of the school day for research purposes only.
    d. Allow the researcher to collect and analyse saliva samples for this research and for potential use in future studies with the researcher permission. I/we understand that my/our son's data will be de-identified.
    e. I/we am/are aware that my son will be identified via a 3-digit code to maintain anonymity.
- I/we am/are aware that all data will be treated with respect, privacy and will be held securely by the schools of Psychology and Biomedical Sciences at Curtin University for at least 5 years then destroyed.
- I/we acknowledge that information generated from this research will retain my/our and my/our son's anonymity and be distributed within Curtin University, academic journals and other venues.
- I/we give consent for the researcher to record information and understand that participation is voluntarily and can be withdrawn at any time without prejudice/penalty.
- I/we acknowledge that I/we can be provided a copy this form, the final report or any other information regarding this study upon request.
- I/we acknowledge that the potential benefits of this research are in excess of the risks of participation.

☐ **YES**, _____ I/we **would like to** register my/our and my/our son's participation for **Study 3** of Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism from 2012-2013.

Child Name: PARTICIPANT NAME

Parent/Primary Carers Name(s): PRIMARY CARER

Parent/Primary Carers Signature(s): _____

Contact Phone Number: PRIMARY CARER TELEPHONE

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

To return this form please:
Scan & email: jeremy.k.marriott@postgrad.curtin.edu.au
or
Fax: (08) 9266 3178
Thank you for your time.

If you require further information please contact Prof Jan Piek (08) 9266 7990 at the School of Psychology and Speech Pathology, Curtin University. If you wish to contact someone independent of the study with any questions or concerns, please contact the Office or Research and Development at Curtin University on (08) 9266 7863.This study has been approved by the Curtin University Human Research Ethics Committee (Approval Number **HR138/2011**). The Committee is comprised of members of the public, academics, lawyers, doctors and pastoral carers. Its main role is to protect participants. If needed, verification of approval can be obtained either by writing to the Curtin University Human Research Ethics Committee, c/- Office of Research and Development, Curtin University, GPO Box U1987, Perth, 6845 or by telephoning (08) 9266 2784 or by emailing hrec@curtin.edu.au

Study 3: School Principal Information Letter.

Curtin University

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

SCHOOL NAME
PRINCIPAL NAME
STREET ADDRESS
SUBURB WA POSTCODE

Dear PRINCIPAL,

Invitation to participate in Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism

The School of Psychology and Speech Pathology and the University Human Research Ethics Committee at Curtin University along with the Department of Education and the Public Transport Authority WA – School Bus Services have approved the above mentioned investigation. This doctoral research is designed to assess the potential for listening to calming music to reduce arousal and Self-Injurious Behaviour (SIB) for males with autism and an intellectual disability (known as Low Functioning Autism or LFA). The potential beneficiaries of this experimental research include people with LFA, parents/primary carers, therapists, teachers and schools.

Reports suggest that up to 40% of people with LFA exhibit SIB. Among school-aged children, this behaviour can result in attention deficits, low academic achievement, limited social development and serious health concerns. Parents, carer givers, teachers and schools are often left searching for treatments to minimise this behaviour other than medication and more invasive procedures. The following 2 studies have emerged from the contemporary literature and my own clinical experience.

Study 3 (registered with the Australian New Zealand Clinical Trials Registry ACTRN12611001064998) 6 school-aged males with LFA who exhibit high frequencies of SIB will listen to calming music through a speaker for the final 14 minutes of their usual morning bus/vehicle journey to school. Saliva will be collected then analysed for the hormone Cortisol and enzyme Alpha Amylase (biological markers of arousal). In addition, usually occurring SIB *attempts* will be analysed via video footage taken (exclusively of the participant and for research purposes only) on the way to school via bus/car and for the first 20 minutes of the school day. No harm will come to participants as they will be required to wear any usual personal protective equipment. I will collect of 5 saliva samples and record footage once per week over 4 consecutive weeks.

Your school is invited to consent to the distribution of an Information Letter and Consent Form to the parents of potential participants and to enable school staff to collaborate with the researcher in the collection of saliva samples.

Please know that all information collected for this research is de-identified, anonymous and kept securely in confidence. We acknowledge that your participation is voluntary and uphold your right to withdraw at any time without prejudice/penalty. To participate, please complete the attached ***School Consent*** fax. Thank you for your time.

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

Study 3: School Principal Consent Form.

Curtin University
School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

Attention	PRINCIPAL NAME
School	SCHOOL NAME
From	Jeremy Marriott
Subject	Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism.
Telephone	0412 320 762
Facsimile	(08) 9266 3178
Number of pages	1

SCHOOL PARTICIPATION

- I have received the information letter and understand the purpose and procedures of *Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism*.
- I understand that the school's involvement is voluntary and that withdrawal is supported at any time without prejudice/penalty.
- I understand that all information will be de-identified, confidential and stored securely at Curtin University for 5 years before being destroyed. I acknowledge that this de-identified information will be distributed to Curtin University, academic journals and other appropriate venues.
- I understand that the school's contribution and identity will not be revealed without written consent.
- I understand that that researcher has a current National Police Certificate and Working with Children Card.
- I am aware that the researcher will access school grounds when required for sampling and video recording of the participant only.
- I acknowledge that the school can be provided a copy this form upon request.
- I have been given the opportunity to ask questions.

☐ **YES**, _____ (School name) **would like to** register our participation for Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism from 2012-2013.

Principal Name: _____

Principal Signature: _____

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

To return this form please:

Scan & email: jeremy.k.marriott@postgrad.curtin.edu.au
or
Fax: (08) 9266 3178

Thank you for your time.

Study 3: School Classroom Teacher Information Letter.

Curtin University

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

Dear TEACHER,

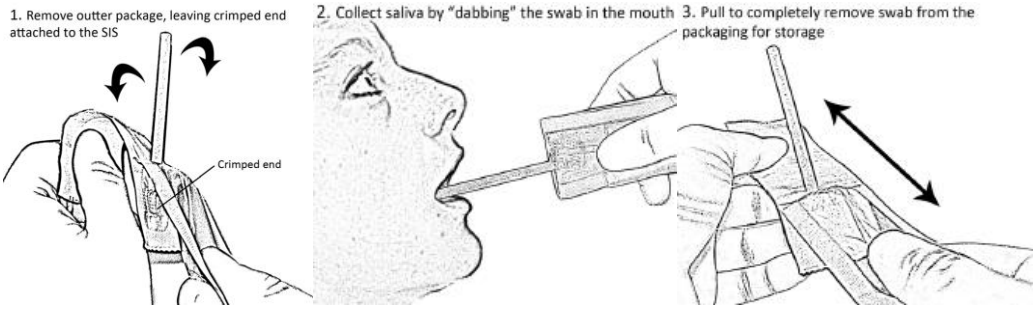
The School of Psychology and Speech Pathology and the University Human Research Ethics Committee at Curtin University along with the Department of Education, Australian New Zealand Clinical Trials Registry and the Public Transport Authority School Bus Services have endorsed an investigation into the potential for calming music listening to reduce arousal and Self-Injurious Behaviour (Mesibov) for boys with autism and an intellectual disability. SCHOOL PRINCIPAL of SCHOOL has agreed to participate in this doctoral research entitled *Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism*. This research is funded by Curtin University and the Health Department WA.

Current treatments designed to reduce SIBs include medication and/or more invasive procedures. The rationale for this research has emerged from a thorough literature review which revealed that music has the potential to reduce arousal and that reducing arousal has the potential to reduce SIB. We invite you to participate in Study 3 as the final stage of investigating this rationale.

Study 3: 6 school-aged boys will listen to music for the last 14 minutes of their morning journey to school. I will use a Salimetrics Children's Swab (SCS) to collect small saliva samples and analyse them for 3 biomarkers of arousal. In addition, I will record footage (exclusively of the participant and for research purposes only) both on their way to school and for the first 20 minutes of their school day. The saliva swabs have been specifically designed to taste pleasant and present no choking hazard (Salimetrics, 2011, p. 8-9):

Instructions for Use – The complete *Salimetrics Saliva Collection and Handling Advice* (2011) will be provided if required

1. For the Salimetrics Children's Swab, peel open the outer package and remove the device ... but leave the crimped end of the swab attached to the packaging during collection, in order to prevent a choking hazard.
2. Securely hold one end of the device and try to place the other end under the child's tongue ... it may only be possible to collect pooling saliva (often at the corners of the mouth). Re-introduce into the mouth as needed until the lower third of the swab is saturated (60-90 seconds total).
3. Fully peel back the outer package to remove the swab for storage.



The saliva samples (and footage) will be collected before, immediately after, 5,10 and 20 minutes after arriving at school then analysed for the hormone Cortisol and enzyme Alpha Amylase as biomarkers of arousal one day each week over 4 consecutive weeks.

Your participation, although voluntary, will play a major role in the assessment of this innovative, safe and non-invasive experimental treatment. Please know that your contribution will remain anonymous, confidential and we support your withdrawal at any time without prejudice/penalty. Your school will receive a summary report of the research results which will be made available to you. If you can participate, please complete the attached form and fax it back then we can provide more detailed information if required then seek final confirmation. Please contact me if you have any questions or if I can provide any further information.

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

Study 3: School Classroom Teacher Consent Form.

Curtin University

10 October 2013

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

I _____ of **SCHOOL** agree to participate in Study 3 of the experimental research entitled: **Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism** conducted by Jeremy Marriott of Curtin University.
I/we give written consent that:

- I have received an information letter and understand the purpose and procedures of Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism.
- I have been given the opportunity to ask and have questions answered prior to consenting.
- I understand that the researcher has a National Police Certificate & Working with Children Card.
- I understand that my involvement will be confidential.
- I understand the importance of the participant not deviating from regular schooling for the duration of the Study 3.
- I am aware that I may be required to assist with:
    a. Collection of saliva samples and recording footage over 4 consecutive weeks in collaboration with the researcher.
    b. I am aware that the saliva sample data from this research may be used in future studies with researcher permission. I am also aware that my identity and that of the saliva donor will be de-identified and remain anonymous.
    c. Video footage will be used for research purposes only and destroyed after being held securely for 5 years.
    d. I am aware that the student will be identified with a 3-digit code to maintain anonymity.
- I am aware that all data will be treated with respect, privacy and held securely by the schools of Psychology and Biomedical Sciences at Curtin University for at least 5 years then be destroyed.
- I acknowledge that information generated as a result of the research will retain my anonymity and be distributed to Curtin University, academic journals and other venues.
- I give consent for the researcher to record information and understand that participation is voluntarily and can be withdrawn at any time without prejudice/penalty.
- I acknowledge that I can be provided a copy this form, the final report or any other information regarding this study upon request.
- I acknowledge that the potential benefits of this research are in excess of the risks of participation.

☐ **YES**, _____ I **would like to** register my participation for Study 3 Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism from 2012-2013.

Teacher Name: _____

Teacher Signature: _____

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

To return this form please:

Scan & email: jeremy.k.marriott@postgrad.curtin.edu.au
or
Fax: (08) 9266 3178

Thank you for your time.

Study 3: School Bus Driver Information Letter.

Curtin University

School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

SCHOOL BUS CONTRACTOR
COMPANY
Street Address
Suburb WA Postcode

Dear SCHOOL BUS CONTRACTOR,

Invitation to participate in Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism

The School of Psychology and Speech Pathology and the University Human Research Ethics Committee at Curtin University, Department of Education, Australian New Zealand Clinical Trials Registry and the Public Transport Authority – School Bus Services have endorsed the above mentioned doctoral research. This study assesses the potential for listening to calming music to reduce salivary biomarkers of arousal and Self-Injurious Behaviour (SIB) for boys with autism and an intellectual disability (known as Low Functioning Autism or LFA). Potential beneficiaries include students with LFA, parents/primary carers, therapists, teachers, schools and school bus drivers.

Reports suggest that up to 40% of people with LFA exhibit SIB. Among the school-aged, this can result in attention deficits, low academic achievement, limited social development and serious health concerns. Unfortunately, the long term prognosis for these children is grim. Families, direct carers, schools and their teachers are often left searching for an effective treatment for SIB. Current treatments include medication and/or more invasive procedures. The purpose of this research is to assess the potential of music to reduce arousal and if reducing arousal can reduce SIB.

School-aged boys with LFA and SIB will listen to calming piano music during their usual journey to school. Saliva samples will be collected immediately before, after, 5, 10 and 20 minutes after the journey to school has concluded. Samples will then be analysed for 3 biological markers of arousal. In addition, footage of usually occurring *SIB attempts* will be recorded along the way. The phrase *SIB attempts* indicates that boys will wear usual personal protective equipment at all times to ensure no harm.

This study will run for 1 school morning each week over 4 consecutive weeks. No music days involve collecting saliva samples and recording SIB attempts for the last 14 minutes of the ride to school. Music days' mirror those of no music days, with one difference. Calming music will be played through a wireless speaker (affixed under participant's seat) for the 14 minutes. At any stage, should the participant's behaviour contravene the Public Transport Authority's behaviour policy, the researcher will cease the treatment.

Participating schools, parents/primary carers, school staff and transport providers will be given information sheets and asked to provide informed consent prior to the commencement of this study. Importantly the researcher will be present for all treatments and all information will be de-identified, anonymous and confidential. Further, it is acknowledged that participation is voluntary and withdrawal at any time without prejudice/penalty is supported. To participate, please complete the attached *Diver Consent* fax. Thank you for your time, and please contact me directly for further information if required.

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

Study 3: School Bus Driver Consent Form.

Curtin University
School of Psychology and Speech Pathology
GPO Box U1987
Perth Western Australia 6845
Telephone +61 8 9266 7279
Facsimile +61 8 9266 2464
Email psych-office@exchange.curtin.edu.au
Web psych.curtin.edu.au

10 October 2013

Attention	SCHOOL BUS CONTRACTOR
Company	SCHOOL BUS CONTRACTOR COMPANY
From	Jeremy Marriott
Subject	Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism.
Telephone	0412 320 762
Facsimile	(08) 9266 3178
Number of pages	1

SCHOOL PARTICIPATION

- I have received an information letter and understand the purpose and procedures of Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism – Study 3.
- I understand that my involvement is voluntary and that withdrawal is supported at any time without prejudice/penalty.
- I understand that all information will be de-identified, confidential and stored securely at Curtin University for 5 years before being destroyed. I acknowledge that information will be distributed to Curtin University, academic journals and other appropriate venues.
- I understand that my contribution and identity will not be revealed without written consent.
- I understand that the researcher has a current National Police Certificate and Working with Children Card.
- I understand that footage will be recorded and saliva samples collected in the vehicle.
- I am aware that the researcher will access the vehicle once each week over 4 consecutive weeks.
- I acknowledge that I can be provided a copy this form upon request.
- I have been given the opportunity to ask questions.

☐ **YES**, _____ (Driver) **would like to** register participation for Music, Arousal and Self-Injurious Behaviour: A three-stage mediating model for Children with Low Functioning Autism from 2012-2013.

Driver Name: _____

Driver Signature: _____

Kind Regards,

Jeremy Marriott
Curtin University | Clinical Psychology PhD Candidate
Department of Psychology & Speech Pathology
HREC | HR138/2011
Study 2 | ACTRN12611001063909
Study 3 | ACTRN12611001064998
Mobile | 0412 320 762
Email | jeremy.k.marriott@postgrad.curtin.edu.au

To return this form please:

Scan & email: jeremy.k.marriott@postgrad.curtin.edu.au
or
Fax: (08) 9266 3178

Thank you for your time.

Appendix T. Study 3: Clinical Interview Protocol Topics (Asperger, 1944 as cited in Frith, 1991; Kanner, 1943).

Autistic psychopathy' in childhood (Asperger, 1944 as cited in Frith, 1991, pp. 39-50) Case: *Fritz V.*	*Autistic disturbances of affective contact* (Kanner, 1971, pp. 217-222) Case: *Donald T.*
General	**General**
1) Birth	1) Birth
2) Age of parents at birth	2) Suckling
3) Familial characteristics in detail	3) Food intake
4) Origin of family	4) Age first talked
5) Fritz V's ASD as defined by parents	5) Age first walked
6) Immediate family structure	6) Developmental milestones met or unmet
7) Family dynamics	7) Speech and language development
8) Developmental milestones and deficits	8) Parental observations
9) Age first walked	9) Compliance to task commands
10) Gross motor skills	10) Environmental exposure
11) Fine motor skills	11) Parental descriptions
12) Practical routines: washing, dressing, cleaning, social skills, eating, attention, could be described as	12) Ages, occupations and descriptions of immediate family
13) Language development	13) School behaviour, independence and academic performance
14) First word spoken at what age	**ASD features:**
15) Childhood illnesses, brain insults or diseases	1) Special interest areas
16) Compliance to parental instruction	2) Attachment style
17) Temperament	3) Social behaviour and abilities
18) Social abilities	4) Sensory and tactile aversions
19) Social interests	5) Repetitive movements
20) Aggressive behaviour	6) Demeanour
21) School behaviour	7) Donald T's ASD as defined by parents
22) Affect	8) Stock social and repetitive phrases
23) Stereotypic movements	9) Communicative abilities
	10) Rituals
Appearance and expressive characteristics:	11) Direct eye to eye gaze
1) Physical description	
2) Height	
3) Skin complexion	
4) Posture	
5) Eye contact, social eye contact	
6) Description of voice	
7) Rhythm and speed of speech	
8) Speech content	

9) Verbal expression 10) Sensory processing preferences 11) Demeanour **Behaviour:** 1) Awareness of surroundings and others 2) Interaction with same-aged peers 3) Inappropriate use of objects 4) Sensory preferences 5) Response to the approach of others 6) Stereotypic movements 7) Precipitants to behaviour 8) Attentional capacity 9) Challenging behaviours 10) Emotional affect 11) Facial expression description 12) Impacts of behaviour on others 13) Response to task commands 14) Individual and group behaviour 15) Fine and gross motor abilities 16) Spatial awareness 17) Intellectual/cognitive ability 18) Consistency of social response 19) Non-verbal communication 20) Special interest areas 21) Special abilities 22) Communication preferences 23) Emotion expression 24) Learning difficulties 25) Demeanour **Differential diagnosis** 1) Comorbidities or differential diagnoses 2) Personality type 3) Overall clinical impression	

Appendix U. Study 3: Fisher's Exact 2 x 3 cell counts and frequencies regarding the tests for an association between sCort concentration and SIB frequencies for Bill, Harry and George.

Study 3: Bill, Harry and George's sCort * SIB Cross tabulation

			SIB			
			0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 Intervals)	Total
Bill's sCort	Below the mean	Count	2	2	5	9
		% within sCort	22.2%	22.2%	55.6%	100.0%
		% within SIB	33.3%	100.0%	50.0%	50.0%
		% of Total	11.1%	11.1%	27.8%	50.0%
	Above The mean	Count	4	0	5	9
		% within sCort	44.4%	0.0%	55.6%	100.0%
		% within SIB	66.7%	0.0%	50.0%	50.0%
		% of Total	22.2%	0.0%	27.8%	50.0%
	Total	Count	6	2	10	18
		% within sCort	33.3%	11.1%	55.6%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	33.3%	11.1%	55.6%	100.0%
Harry's sCort	Below the mean	Count	0	3	4	7
		% within sCort	0.0%	42.9%	57.1%	100.0%
		% within SIB	0.0%	60.0%	50.0%	46.7%
		% of Total	0.0%	20.0%	26.7%	46.7%
	Above The mean	Count	2	2	4	8
		% within sCort	25.0%	25.0%	50.0%	100.0%
		% within SIB	100.0%	40.0%	50.0%	53.3%
		% of Total	13.3%	13.3%	26.7%	53.3%
	Total	Count	2	5	8	15
		% within sCort	13.3%	33.3%	53.3%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	13.3%	33.3%	53.3%	100.0%
George's sCort	Below the mean	Count	7	1	4	12
		% within sCort	58.3%	8.3%	33.3%	100.0%
		% within SIB	77.8%	50.0%	44.4%	60.0%
		% of Total	35.0%	5.0%	20.0%	60.0%
	Above The mean	Count	2	1	5	8
		% within sCort	25.0%	12.5%	62.5%	100.0%
		% within SIB	22.2%	50.0%	55.6%	40.0%
		% of Total	10.0%	5.0%	25.0%	40.0%
	Total	Count	9	2	9	20
		% within sCort	45.0%	10.0%	45.0%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	45.0%	10.0%	45.0%	100.0%

Study 3: Fisher's Exact 2 x 3 cell counts and frequencies regarding the tests for an association between sAA activity and SIB frequencies for Bill, Harry and George.

Study 3: Bill, Harry and George's sAA * SIB Cross tabulation

			SIB			
			0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 intervals)	Total
Bill's sAA	Below the mean	Count	4	1	5	10
		% within sAA	40.0%	10.0%	50.0%	100.0%
		% within SIB	66.7%	50.0%	41.7%	50.0%
		% of Total	20.0%	5.0%	25.0%	50.0%
	Above The mean	Count	2	1	7	10
		% within sAA	20.0%	10.0%	70.0%	100.0%
		% within SIB	33.3%	50.0%	58.3%	50.0%
		% of Total	10.0%	5.0%	35.0%	50.0%
	Total	Count	6	2	12	20
		% within sAA	30.0%	10.0%	60.0%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	30.0%	10.0%	60.0%	100.0%
Harry's sAA	Below the mean	Count	1	4	5	10
		% within sAA	10.0%	40.0%	50.0%	100.0%
		% within SIB	50.0%	50.0%	55.6%	52.6%
		% of Total	5.3%	21.1%	26.3%	52.6%
	Above The mean	Count	1	4	4	9
		% within sAA	11.1%	44.4%	44.4%	100.0%
		% within SIB	50.0%	50.0%	44.4%	47.4%
		% of Total	5.3%	21.1%	21.1%	47.4%
	Total	Count	2	8	9	19
		% within sAA	10.5%	42.1%	47.4%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	10.5%	42.1%	47.4%	100.0%
George's sAA	Below the mean	Count	5	2	3	10
		% within sAA	50.0%	20.0%	30.0%	100.0%
		% within SIB	55.6%	100.0%	33.3%	50.0%
		% of Total	25.0%	10.0%	15.0%	50.0%
	Above The mean	Count	4	0	6	10
		% within sAA	40.0%	0.0%	60.0%	100.0%
		% within SIB	44.4%	0.0%	66.7%	50.0%
		% of Total	20.0%	0.0%	30.0%	50.0%
	Total	Count	9	2	9	20
		% within sAA	45.0%	10.0%	45.0%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	45.0%	10.0%	45.0%	100.0%

Study 3: Fisher's Exact 3 x 3 cell counts and frequencies regarding the tests for an association between joint sCort concentration and sAA activity and SIB frequencies for Bill, Harry and George.

Joint sCort/sAA * Behaviour Cross tabulation

			SIB			
			0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 intervals)	Total
Bill's Joint sCort/sAA	Both sCort and sAA above the mean	Count	1	0	3	4
		% within Joint sCort/sAA	25.0%	0.0%	75.0%	100.0%
		% within SIB	16.7%	0.0%	30.0%	22.2%
		% of Total	5.6%	0.0%	16.7%	22.2%
	Both sCort and sAA below the mean	Count	1	1	2	4
		% within Joint sCort/sAA	25.0%	25.0%	50.0%	100.0%
		% within SIB	16.7%	50.0%	20.0%	22.2%
		% of Total	5.6%	5.6%	11.1%	22.2%
	sCort above and sAA below the mean	Count	3	0	2	5
		% within Joint sCort/sAA	60.0%	0.0%	40.0%	100.0%
		% within SIB	50.0%	0.0%	20.0%	27.8%
		% of Total	16.7%	0.0%	11.1%	27.8%
	sCort below and sAA above the mean	Count	1	1	3	5
		% within Joint sCort/sAA	20.0%	20.0%	60.0%	100.0%
		% within SIB	16.7%	50.0%	30.0%	27.8%
		% of Total	5.6%	5.6%	16.7%	27.8%
	Total	Count	6	2	10	18
		% within Joint sCort/sAA	33.3%	11.1%	55.6%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	33.3%	11.1%	55.6%	100.0%
Harry's Joint sCort/sAA	Both sCort and sAA above the mean	Count	1	1	1	3
		% within Joint sCort/sAA	33.3%	33.3%	33.3%	100.0%
		% within SIB	50.0%	20.0%	12.5%	20.0%
		% of Total	6.7%	6.7%	6.7%	20.0%
	Both sCort and sAA below the mean	Count	0	2	2	4
		% within Joint sCort/sAA	0.0%	50.0%	50.0%	100.0%
		% within SIB	0.0%	40.0%	25.0%	26.7%
		% of Total	0.0%	13.3%	13.3%	26.7%
	sCort above and sAA below the mean	Count	1	1	3	5
		% within Joint sCort/sAA	20.0%	20.0%	60.0%	100.0%
		% within SIB	50.0%	20.0%	37.5%	33.3%
		% of Total	6.7%	6.7%	20.0%	33.3%
	sCort below and sAA above the mean	Count	0	1	2	3
		% within Joint sCort/sAA	0.0%	33.3%	66.7%	100.0%
		% within SIB	0.0%	20.0%	25.0%	20.0%
		% of Total	0.0%	6.7%	13.3%	20.0%
	Total	Count	2	5	8	15
		% within Joint sCort/sAA	13.3%	33.3%	53.3%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	13.3%	33.3%	53.3%	100.0%

George's Joint sCort/sAA	Both sCort and sAA above the mean	Count	1	0	3	4
		% within Joint sCort/sAA	25.0%	0.0%	75.0%	100.0%
		% within SIB	11.1%	0.0%	33.3%	20.0%
		% of Total	5.0%	0.0%	15.0%	20.0%
	Both sCort and sAA below the mean	Count	4	1	1	6
		% within Joint sCort/sAA	66.7%	16.7%	16.7%	100.0%
		% within SIB	44.4%	50.0%	11.1%	30.0%
		% of Total	20.0%	5.0%	5.0%	30.0%
	sCort above and sAA below the mean	Count	1	1	2	4
		% within Joint sCort/sAA	25.0%	25.0%	50.0%	100.0%
		% within SIB	11.1%	50.0%	22.2%	20.0%
		% of Total	5.0%	5.0%	10.0%	20.0%
	sCort below and sAA above the mean	Count	3	0	3	6
		% within Joint sCort/sAA	50.0%	0.0%	50.0%	100.0%
		% within SIB	33.3%	0.0%	33.3%	30.0%
		% of Total	15.0%	0.0%	15.0%	30.0%
	Total	Count	9	2	9	20
		% within Joint sCort/sAA	45.0%	10.0%	45.0%	100.0%
		% within SIB	100.0%	100.0%	100.0%	100.0%
		% of Total	45.0%	10.0%	45.0%	100.0%

Study 3: Fisher's Exact 2 x 2 cell counts and frequencies regarding the tests for an association between sCort concentration and sAA activity for Bill, Harry and George.

Study 3: Bill, Harry and George's sCort * sAA Cross tabulation

			sAA		
			Below the mean	Above the mean	Total
Bill's sCort	Below the mean	Count	4	5	9
		% within sCort	44.4%	55.6%	100.0%
		% within sAA	44.4%	55.6%	50.0%
		% of Total	22.2%	27.8%	50.0%
	Above The mean	Count	5	4	9
		% within sCort	55.6%	44.4%	100.0%
		% within sAA	55.6%	44.4%	50.0%
		% of Total	27.8%	22.2%	50.0%
	Total	Count	9	9	18
		% within sCort	50.0%	50.0%	100.0%
		% within sAA	100.0%	100.0%	100.0%
		% of Total	50.0%	50.0%	100.0%
Harry's sCort	Below the mean	Count	4	3	7
		% within sCort	57.1%	42.9%	100.0%
		% within sAA	44.4%	50.0%	46.7%
		% of Total	26.7%	20.0%	46.7%
	Above The mean	Count	5	3	8
		% within sCort	62.5%	37.5%	100.0%
		% within sAA	55.6%	50.0%	53.3%
		% of Total	33.3%	20.0%	53.3%
	Total	Count	9	6	15
		% within sCort	60.0%	40.0%	100.0%
		% within sAA	100.0%	100.0%	100.0%

		% of Total	60.0%	40.0%	100.0%
George's sCort	Below the mean	Count	6	6	12
		% within sCort	50.0%	50.0%	100.0%
		% within sAA	60.0%	60.0%	60.0%
		% of Total	30.0%	30.0%	60.0%
	Above The mean	Count	4	4	8
		% within sCort	50.0%	50.0%	100.0%
		% within sAA	40.0%	40.0%	40.0%
		% of Total	20.0%	20.0%	40.0%
	Total	Total	10	10	20
		% within sCort	50.0%	50.0%	100.0%
		% within sAA	100.0%	100.0%	100.0%
		% of Total	50.0%	50.0%	100.0%

Appendix V. Study 3: Fisher's Exact 2 x 3 cell counts and frequencies regarding the associations between the categorical SIB frequency variable (1 interval, 1-6 intervals, 6 intervals) and the binary 'baseline versus music' variable indicated a significant association between them for Bill.

Study 3: Bill – Condition * SIB frequencies Day 2 Cross tabulation

		Frequency Phase B1			
PIR Obs.		0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 intervals)	Total
0-10mins on the bus before music	Count	8	2	0	10
	% within Condition	80.0%	20.0%	0.0%	100.0%
	% within SIB frequencies Day 2	80.0%	66.7%	0.0%	40.0%
	% of Total	32.0%	8.0%	0.0%	40.0%
11-25mins on the bus during music listening	Count	2	1	12	15
	% within Condition	13.3%	6.7%	80.0%	100.0%
	% within SIB frequencies Day 2	20.0%	33.3%	100.0%	60.0%
	% of Total	8.0%	4.0%	48.0%	60.0%
Total	Count	10	3	12	25
	% within Condition	40.0%	12.0%	48.0%	100.0%
	% within SIB frequencies Day 2	100.0%	100.0%	100.0%	100.0%
	% of Total	40.0%	12.0%	48.0%	100.0%

Study 3: Fisher's Exact 2 x 3 cell counts and frequencies regarding the associations between the categorical SIB frequency variable (1 interval, 1-6 intervals, 6 intervals) and the binary 'baseline versus music' variable indicated a significant association between them for Harry.

Study 3: Harry – Condition * SIB frequencies Day 4 Cross tabulation

		Frequency Phase B2			
PIR Obs.		0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 intervals)	Total
0-10mins on the bus before music	Count	8	1	1	10
	% within Condition	80.0%	10.0%	10.0%	100.0%
	% within SIB frequencies Day 4	80.0%	14.3%	12.5%	40.0%
	% of Total	32.0%	4.0%	4.0%	40.0%
11-25mins on the bus during music listening	Count	2	6	7	15
	% within Condition	13.3%	40.0%	46.7%	100.0%
	% within SIB frequencies Day 4	20.0%	85.7%	87.5%	60.0%
	% of Total	8.0%	24.0%	28.0%	60.0%
Total	Count	10	7	8	25
	% within Condition	40.0%	28.0%	32.0%	100.0%
	% within SIB frequencies Day 4	100.0%	100.0%	100.0%	100.0%
	% of Total	40.0%	28.0%	32.0%	100.0%

Study 3: Fisher's Exact 2 x 3 cell counts and frequencies regarding the associations between the categorical SIB frequency variable (1 interval, 1-6 intervals, 6 intervals) and the binary 'baseline versus music' variable indicated a significant association between them for George on Day 2 (Music).

Study 3: George – Condition * SIB frequencies Day 2 Cross tabulation

PIR Obs.		Frequency Phase B1			Total
		0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 intervals)	
0-10mins on the bus before music	Count	0.0%	0	10	10
	% within Condition	0.0%	0.0%	100.0%	100.0%
	% within SIB frequencies Day 2	0.0%	0.0%	100.0%	40.0%
	% of Total	0.0%	0.0%	40.0%	40.0%
11-25mins on the bus during music listening	Count	13	2	0	15
	% within Condition	86.7%	13.3%	0.0%	100.0%
	% within SIB frequencies Day 2	100.0%	100.0%	0.0%	60.0%
	% of Total	52.0%	8.0%	0.0%	60.0%
Total	Count	13	2	10	25
	% within Condition	52.0%	8.0%	40.0%	100.0%
	% within SIB frequencies Day 2	100.0%	100.0%	100.0%	100.0%
	% of Total	52.0%	8.0%	40.0%	100.0%

Study 3: Fisher's Exact 2 x 3 cell counts and frequencies regarding the associations between the categorical SIB frequency variable (1 interval, 1-6 intervals, 6 intervals) and the binary 'baseline versus music' variable indicated a significant association between them for George on Day 4 (Music).

Study 3: George – Condition * SIB frequencies Day 4 Cross tabulation

PIR Obs.		Frequency Phase B2			Total
		0% (1 Interval)	0% – 99% (1-6 Intervals)	100% (6 intervals)	
0-10mins on the bus before music	Count	0.0%	0	10	10
	% within Condition	0.0%	0.0%	100.0%	100.0%
	% within SIB frequencies Day 4	0.0%	0.0%	43.5%	40.0%
	% of Total	0.0%	0.0%	40.0%	40.0%
11-25mins on the bus during music listening	Count	13	2	13	15
	% within Condition	86.7%	13.3%	86.7%	100.0%
	% within SIB frequencies Day 4	100.0%	100.0%	56.5%	60.0%
	% of Total	52.0%	8.0%	52.0%	60.0%
Total	Count	13	2	23	25
	% within Condition	52.0%	8.0%	92.0%	100.0%
	% within SIB frequencies Day 4	100.0%	100.0%	100.0%	100.0%
	% of Total	52.0%	8.0%	92.0%	100.0%

Appendix W. Bill's Motivation Assessment Scale (MAS) Protocol and Results.

Item	MAS Question	Response
1	Would the behavior occur continuously, over and over, if this person left alone for long periods of time? (For example, several hours.)	6 Always
2	Does the behavior occur following a request to perform a difficult task?	0 Never
3	Does the behavior seem to occur in response to your talking to other person in the room?	0 Never
4	Does the behavior ever occur to get a toy, food or activity that this person has been told that he or she can't have?	0 Never
5	Would the behavior occur repeatedly, in the same way, for very long periods of time, if no one was around? (For example, rocking back and forth for over an hour.)	6 Always
6	Does the behavior occur when any request is made of this person?	0 Never
7	Does the behavior occur when you stop attending to this person?	0 Never
8	Does the behavior occur when you take away a favourite toy, food, or activity?	0 Never
9	Does it appear to you that this person enjoys performing the behavior? (It feels, tastes, looks, smells, and/or sounds pleasing.)	5 Almost Always
10	Does this person seem to do the behavior to upset or annoy you when you are trying to get him or her to do what you ask?	3 Half the Time
11	Does this person seem to do the behavior to upset or annoy you when you are not paying attention to him or her? (For example, if you are sitting in a separate room, interacting with another person.)	0 Never
12	Does the behavior stop occurring shortly after you give this person the toy, food or activity he or she has requested?	0 Never
13	When the behavior is occurring, does this person seem calm and unaware of anything else going on around him or her?	1 Almost Never
14	Does the behavior stop occurring shortly after (one to five minutes) you stop working or making demands of this person?	0 Never
15	Does this person seem to do the behavior to get you to spend some time with him or her?	0 Never
16	Does this behavior seem to occur when this person has been told that he or she can't do something he or she had wanted to do?	0 Never

Domain =	Sensory	Escape	Attention	Tangible
	Item 1. = 6	Item 2. = 0	Item 3. = 0	Item 4. = 0
	Item 5. = 6	Item 6. = 0	Item 7. = 0	Item 8. = 0
	Item 9. = 5	Item 10. = 3	Item 11. = 0	Item 12. = 0
	Item 13. = 1	Item 14. = 0	Item 15. = 0	Item 16. = 0
Total Score =	18	3	0	0
Mean Score =	4.5	0.75	0	0
Rel. Ranking =	1	2	3	3

Appendix X. Study 3: Inter-raters A and B, self-reported qualifications/experience.

Inter-rater A		
Topic	Year	Description
Academic	2015	Curtin University – BSc. Psychology
	2013	ABA 4 Autism – Level 1 Applied Behaviour Analysis
	2012	Academic Events Co-ordinator | Curtin University Psychology Student Council (PSC) This position is responsible for developing and implementing extracurricular academic events for undergraduate Psychology students at Curtin. Responsible for collaborating with service providers, government and community.
	2004	West Cost Institute of Training – Diploma Community Services, Youth Work
Experience	2013	Community Support Officer – Private This role is responsible for supporting children, teenagers and adults liven with mental illness, disability and ASD. As a participant driven program my position involves the facilitation of activities aimed at increasing independence and life-long learning.
	2013	Applied Behaviour Analysis Therapist: I am responsible for providing intensive home based ABA to children with ASD, shape and teach behaviours.
Inter-rater B		
Topic	Year	Description
Academic	2015	Curtin University – BSc. Psychology (Hons)
	2016	Curtin University – BA. Psychology
Experience	2015	I am now working as a developmental therapist with children with autism. At the moment I am working with 7 children of different levels of needs, mostly high functioning but I have just started working with a low functioning 4-year-old. (The kids range from age 4-6 as it is early intervention). Just a little bit of feedback from my kids – I have been told I am gorgeous, I have been told "I love you" I have been asked to give cuddles, I have been invited to birthday parties, the beach, shopping, swimming and have played with teddies even mum is not allowed to touch! I feel I have a connection with children with autism, many people think my job is challenging – and whilst it can be at times, it is really rewarding watching the children grow, learn and develop with things I am teaching them, I love my job! Furthermore, I have applied for my general registration as a psychologist. Currently keeping fingers crossed that AHPRA are impressed with my 30+ page application! Outside of this I have many books on autism and have read many true stories written about learning about the development of children with autism, how to work with them, how to gain their interest.

Appendix Y. Harry and George's Motivation Assessment Scale (MAS) Protocol and Results.

Item	MAS Question	Response
1	Would the behavior occur continuously, over and over, if this person left alone for long periods of time? (For example, several hours.)	5 Almost Always
2	Does the behavior occur following a request to perform a difficult task?	0 Never
3	Does the behavior seem to occur in response to your talking to other person in the room?	2 Seldom
4	Does the behavior ever occur to get a toy, food or activity that this person has been told that he or she can't have?	0 Never
5	Would the behavior occur repeatedly, in the same way, for very long periods of time, if no one was around? (For example, rocking back and forth for over an hour.)	6 Always
6	Does the behavior occur when any request is made of this person?	3 Half the Time
7	Does the behavior occur when you stop attending to this person?	0 Never
8	Does the behavior occur when you take away a favourite toy, food, or activity?	6 Always
9	Does it appear to you that this person enjoys performing the behavior? (It feels, tastes, looks, smells, and/or sounds pleasing.)	4 Usually
10	Does this person seem to do the behavior to upset or annoy you when you are trying to get him or her to do what you ask?	0 Never
11	Does this person seem to do the behavior to upset or annoy you when you are not paying attention to him or her? (For example, if you are sitting in a separate room, interacting with another person.)	0 Never
12	Does the behavior stop occurring shortly after you give this person the toy, food or activity he or she has requested?	6 Always
13	When the behavior is occurring, does this person seem calm and unaware of anything else going on around him or her?	0 Never
14	Does the behavior stop occurring shortly after (one to five minutes) you stop working or making demands of this person?	0 Never
15	Does this person seem to do the behavior to get you to spend some time with him or her?	0 Never
16	Does this behavior seem to occur when this person has been told that he or she can't do something he or she had wanted to do?	2 Seldom

Domain =	Sensory	Escape	Attention	Tangible
	Item 1. = 5	Item 2. = 0	Item 3. = 2	Item 4. = 0
	Item 5. = 6	Item 6. = 3	Item 7. = 0	Item 8. = 6
	Item 9. = 4	Item 10. = 0	Item 11. = 0	Item 12. = 6
	Item 13. = 0	Item 14. = 0	Item 15. = 0	Item 16. = 2
Total Score =	15	3	2	14
Mean Score =	3.75	0.75	0.5	3.5
Rel. Ranking =	1	3	4	2

George's Motivation Assessment Scale (MAS) Protocol and Results.

Item	MAS Question	Response
1	Would the behavior occur continuously, over and over, if this person left alone for long periods of time? (For example, several hours.)	5 Almost Always
2	Does the behavior occur following a request to perform a difficult task?	3 Half the Time
3	Does the behavior seem to occur in response to your talking to other person in the room?	3 Half the Time
4	Does the behavior ever occur to get a toy, food or activity that this person has been told that he or she can't have?	4 Usually
5	Would the behavior occur repeatedly, in the same way, for very long periods of time, if no one was around? (For example, rocking back and forth for over an hour.)	6 Always
6	Does the behavior occur when any request is made of this person?	3 Half the Time
7	Does the behavior occur when you stop attending to this person?	5 Almost Always
8	Does the behavior occur when you take away a favourite toy, food, or activity?	5 Almost Always
9	Does it appear to you that this person enjoys performing the behavior? (It feels, tastes, looks, smells, and/or sounds pleasing.)	6 Always
10	Does this person seem to do the behavior to upset or annoy you when you are trying to get him or her to do what you ask?	3 Half the Time
11	Does this person seem to do the behavior to upset or annoy you when you are not paying attention to him or her? (For example, if you are sitting in a separate room, interacting with another person.)	3 Half the Time
12	Does the behavior stop occurring shortly after you give this person the toy, food or activity he or she has requested?	5 Almost Always
13	When the behavior is occurring, does this person seem calm and unaware of anything else going on around him or her?	3 Half the Time
14	Does the behavior stop occurring shortly after (one to five minutes) you stop working or making demands of this person?	2 Seldom
15	Does this person seem to do the behavior to get you to spend some time with him or her?	2 Seldom
16	Does this behavior seem to occur when this person has been told that he or she can't do something he or she had wanted to do?	5 Almost Always

Domain =	Sensory	Escape	Attention	Tangible
	Item 1. = 5	Item 2. = 3	Item 3. = 3	Item 4. = 4
	Item 5. = 6	Item 6. = 3	Item 7. = 5	Item 8. = 5
	Item 9. = 6	Item 10. = 3	Item 11. = 3	Item 12. = 5
	Item 13. = 3	Item 14. = 2	Item 15. = 2	Item 16. = 5
Total Score =	20	11	13	19
Mean Score =	5	2.75	3.25	4.75
Rel. Ranking =	1	4	3	2

Appendix Z. Copyright permissions for Figure 2.2, Figure 2.3 and Table 4.1.

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