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# Restricted and Repetitive Behaviours (RRBs) across Three Emotional Contexts and their relationship with Executive Functioning and Emotion Regulation

Thesis submitted for the degree of Doctor of Philosophy

School of Psychology and Clinical Language Sciences

**University of Reading (Malaysia Campus)**

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# Declaration of original authorship

I confirm that this is my own work and the use of all material from other sources has been properly and fully acknowledged.

Ling Chih Chong

# Acknowledgements

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# Covid-19 Impact Statement

Malaysia was affected by the Covid-19 pandemic, which had a significant impact on the work of this thesis. Malaysia implemented several nationwide "Movement Control Orders" (MCO) to mitigate the spread of Covid-19. It disrupted on-going experiments, limited access to resources and facilities, created challenges in communication and impacted recruitment of research participants.

The first study of this thesis received its ethics approval in February 2020, which was the beginning of the pandemic. This study intended to examine the usability of a measurement tool that was developed in this thesis by collecting real-time data when participants performed Jumping Jacks at the laboratory in University of Reading Malaysia. The timing led to recruitment difficulties.

The ethics approval for Study 2 was given in March 2020. Following a spike in local Covid-19 cases, Malaysia implemented its first MCO. During this period all higher education institutes were closed, and only essential activities, were allowed. Experiments involving in-person implementation and observation were impossible and therefore, had to be put on hold.

Although there had been intentions of easing restrictions, the MCO was reimposed repeatedly from March 18, 2020, until June 14, 2021, due to surges in Covid-19 cases. My supervisors and I had to submit several requests to the University of Reading Malaysia Research Ethics Committee (UoRM REC) to modify the procedures of our studies due to the constant changes. These extenuating circumstances forced our studies to shift the design from in-person neuropsychological testing and behavioural observation to online questionnaires. While acknowledging the limitations of using only questionnaires to answer our research questions, they provided a higher flexibility in implementation, allowing scientific progression even whilst social restrictions were in place.

In January 2022, although certain restrictions such as capacity limits were still in place, on-site and in-person experiments were allowed in controlled circumstances. We gained ethical approval from UoRM REC and the Ministry of Education to carry out our experiments in primary school settings.

# List of Abbreviations

| | |
|---|---|
| ADI-R | Autism Diagnostic Interview-Revised |
| ADOS | Autism Diagnostic Observational Schedule |
| ASD/ASC | Autism Spectrum Disorder/Autism Spectrum Conditions |
| BDI-2 | Battelle Developmental Inventory, Second Edition |
| BISCUIT-Part3 | Baby and Infant Screen for Children with aUtIsm Traits – Part3 |
| BRIEF | Behavior Rating Inventory of Executive Function |
| BRIEF-P | Behavior Rating Inventory of Executive Function-Preschool Version |
| BSID | Bayley Scales of Infant Development |
| CBCL | Child Behavior Checklist |
| CDD | Childhood Disintegrative Disorder |
| CI | Circumscribed Interests |
| DQ | Developmental Quotient |
| DSM | Diagnostic and Statistical Manual of Mental Disorder |
| EFA | Exploratory Factor Analysis |
| HFA | High-Functioning Autism |
| ICD | International Classification of Diseases |
| ID | Intellectual Disabilities |
| IQ | Intelligent Quotient |
| IS | Insistence on Sameness |
| M-CHAT | Modified Checklist for Autism in Toddlers |
| MRI | Magnetic Resonance Imaging |
| MSEL | Mullen Scales of Early Learning |
| NS | Nonspectrum disorder |
| NVIQ | Nonverbal IQ |
| OCD | Obsessive and Compulsive Disorder |
| PCA | Principal Component Analysis |
| PDD | Pervasive Developmental Disorders |
| PDD-NOS | Pervasive Developmental Disorder, Not Otherwise Specified |

| | |
|---|---|
| RBQ | Repetitive Behaviour Questionnaire |
| RBS-R | Repetitive Behavior Scale-Revised |
| RBS-R | Repetitive Behaviors Scale – Revised |
| RRBs | Restricted and Repetitive Behaviours |
| SMB | Sensory Motor Behaviours |
| SQ | Sensory Questionnaire |
| TD | Typically Developing |
| Vineland-ll | Vineland Adaptive Behavior Scales, Second Edi-tion |
| ID | Intellectual Disability |
| WISC-IV | Wechsler Intelligence Scale for Children-Fourth Edition |
| VCI | Verbal Comprehension Index |
| PRI | Perceptual Reasoning Index |
| SRS-2 | Social Responsiveness Scale, Second Edition |
| SCI | Social Communication Index |
| SCARED-C/P | Screen for Childhood Anxiety Related Disorders: Child and Parent Versions |
| IUS-C/P | Intolerance of Uncertainty Scale: Child and Parent Versions |
| AS | Asperger's Syndrome |
| DCCS | Dimensional Change Card Sorting |
| DAS-II | Differential Abilities Scale-second edition |
| SCQ | Social Communication Questionnaire |
| SRS | Social Responsiveness Scale-2 |
| CBCL | Child Behavior Checklist |
| ARS | Anger Rumination Scale |
| RPQ | *Reactive-Proactive Aggression Questionnaire* |
| CSP-2 | Child Sensory Profile 2 |
| D-KEFS | Delis-Kaplan executive Functions scale |
| BASC-2 PRS | Behaviour Assessment System for Children Parent Rating Scale |
| ADHD | Attention-deficit/hyperactivity disorder |
| ERICA | Emotion Regulation Index for Children and Adolescents |
| HIF | How I Feel questionnaire |

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# Abstract

Restricted and Repetitive Behaviours (RRBs) have been postulated to serve as a type of early Emotion Regulation (ER) behaviour as they introduce order and predictability to the child's environment through repetition and consequently reduce anxiety. These behaviours decrease in line with the manifestation and advancement of Executive Functioning (EF). It has also been found that EF training significantly improves children's ability in managing emotion. However, no studies have investigated the two-way relationship between EF, ER and RRBs. In this thesis, we investigated whether ER serves as a mediator in the relationship between EF and RRBs with questionnaires, neuropsychological tests, and behavioural observations across three emotional contexts: Task, Preferred and Neutral. Mediation analysis was used to analyse their relationships. Results showed that children with autism engaged in more RRBs during the Preferred condition whereas typically developing children engaged in more RRBs during the Neutral condition, suggesting that RRBs potentially serve different functions for these two populations. ER was considered as a possible mediator in the relationship between EF and RRBs as its mediating effect was significant in parent-report measures but not neuropsychological and observational measures. We also modified a recently developed pose-estimation technology and developed a novel measurement tool, called OpenPose_Angle, that can automatically detect body parts, place keypoints on body segments, compute and analyse angles. Compared to traditional measurements, OpenPose_Angle could provide kinematic data of systematic and repetitive movements in a less resource-intensive manner, with high sensitivity and specificity. This technology was more likely to generate meaningful data for repetitive hand clapping and hand swinging behaviours, but it is still not ready for random and spontaneous repetitive behaviours displayed by children with autism.

# Contents

# Thesis Outline

The main aim of this thesis is to examine the mediating role of Emotion Regulation on the relationship between Executive Functioning and Restrictive and Repetitive Behaviours (RRBs) across three emotional contexts: Task, Preferred and Neutral. A novel measurement tool, called OpenPose_Angle, was developed in this thesis to provide an automated technology to detect body segments, place keypoints and analyse angles. This tool was used to measure the RRBs of children with autism and typically developing children across the three emotional conditions.

In order to adequately examine this topic in more details, a brief grounding in the relevant literature is essential. The first chapter offers an overview of previous work relevant to Executive Functioning, Emotion Regulation and RRBs in typical development and in autism. We then examine the relationship between these three constructs across emotional contexts using parent-report, neuropsychological and observational measures. Finally, the chapters close with an overview of the findings of the thesis and how they relate to research in relevant areas.

**Chapter 1** introduces Executive Functioning, Emotion Regulation, repetitive behaviours and their relationship in typical development. Then, an overview of these three constructs in autism and their relationship is discussed. The relationship between these three constructs is further supported using neurobiological accounts.

**Chapter 2** describes the first empirical study where 64 children participated in an experiment looking at the role of Emotion Regulation as mediator between Executive Functioning and RRBs based on parental self-report measures such as the Childhood Executive Functioning Inventory (CHEXI; Thorell & Nyberg, 2008), the *Emotion Regulation*

*Checklist (ERC;* Shields & Cicchetti, 1997) and the *Repetitive Behaviour Questionnaire-2 (RBQ-2;* Leekam et al., 2007).

**Chapter 3** investigates the occurrences of RRBs across three emotional conditions: Task, Preferred, and Neutral. Then, it describes a replication of the study described in Chapter 2 to investigate the overall robustness of the patterns of the findings and extend the previous study using neuropsychological behavioural tasks and observational measures to examine whether Emotion Regulation remained as a mediator in the relationship between Executive Functioning and RRBs when different measures are used.

**Chapter 4** examines the usability of OpenPose_Angle in measuring repetitive behaviours in autism. First, the consistency of reproducibility of keypoints, sensitivity and specificity of OpenPose_Angle is examined on systematic and repetitive movements performed by typically developing individuals. Then, the system was used to measure the spontaneous and random repetitive behaviours displayed by children with autism and typically developing children.

**Chapter 5** summarises and discusses our findings: how do RRBs potentially serve different functions for typically developing children and children with autism, and how Emotion Regulation mediate the relationship between Executive Functioning and RRBs.

# 1 Chapter One: General Introduction

## 1.1. Potential Roles of EF and ER on RRBs

A causal relationship between Executive Functioning and Emotion Regulation has recently been suggested by Li et al.'s (2020) study when they found Executive Function training improved children's emotional competence. Many past studies (Willemsen-Swinkels et al., 1998, Hirschler-Guttenberg et al., 2015, Çevikaslan et al., 2014) have also observed a relationship between these two constructs and Restricted and Repetitive Behaviours (RRBs) in autism independently. RRBs have also been suggested serving as a strategy to regulate emotions. However, no studies have looked at the two-way relationship between them, Executive Functioning → Emotion Regulation → RRBs. In the beginning of this chapter, an overview of Executive Functioning, Emotion Regulation and RRBs in typical development will be provided. Then, Autism Spectrum Condition (ASC) will briefly be introduced before we review the three constructs in autistic population. Lastly, the postulated two-way relationship between the three target constructs will be discussed.

## 1.2. Executive Functioning (EF)

The term "Executive Functions" has broadly been used for various hypothesised goal-oriented and higher-order cognitive abilities, including cognitive flexibility, inhibitory control, working memory, planning, attention, and self-regulation. Pribram (1973) was the first to use the term "executive function" to refer to the functions of the frontal cortex, "the frontal cortex appears critically involved in implementing executive programs when these are necessary to maintain brain organisation in the face of insufficient redundancy in input processing and the outcomes of behaviour" (Pribram, 1973). Observation and case studies in frontal lobe lesion led researchers to the conclusion that the frontal lobes play a primary role in higher-order functions (Szczepanski & Knight, 2014). Since the introduction of the term, up to approximately 48 models have been developed to explain Executive Functioning (Goldstein & Naglieri, 2014) but there is not one particular model that has general acceptance in the field.

Various models of Executive Functioning were developed utilizing different theoretical paradigms such as clinical, neurobiological, cognitive and behavioural paradigms. The methodologies in Executive Functioning studies can range from measuring cognitive processes with neuropsychological tasks to measuring the neurobiological processes of performance involving Executive Functioning using neuroimaging techniques. A group of Executive Functioning models of cognitive paradigms focused on individual Executive Functioning processes and these models have reported a number of Executive Functioning constructs ranged from 2 (Miyake & Friedman, 2012) to 30 (Baggetta & Alexander, 2016). In the early research, Stuss and Benson (1986) used Executive Functioning as “a generic term that refers to a variety of different capacities that enable purposeful, goal-directed, including behaviour regulation, working memory, planning and organisational skills and self-monitoring” (p. 272). Ozonoff et al. (1991) wrote that Executive Functioning skills refer to a set of cognitive abilities necessary “to maintain an appropriate problem-solving set for attainment of a future goal” (p. 1083). More recently, Naglieri and Goldstein (2013) put forward the Executive Functioning as an umbrella term referring to the ability to acquire skills across nine areas, including attention, working memory, inhibitory control, cognitive flexibility, emotion regulation, initiation, organisation, planning, and self-monitoring. The three most commonly reported Executive Functioning constructs in literatures are cognitive flexibility, working memory, and response inhibition (Demetriou et al., 2019; Hill, 2004; Memari et al., 2013; Wang et al., 2017).

Cognitive flexibility refers to the ability to switch back and forth between different tasks and behavioural responses (Miyake et al., 2000). It is an important developmental achievement to flexibly control our thoughts, behaviours and emotions, especially when confronted with conflicting desires and habits. Children with poor cognitive flexibility are more likely to have difficulties in simple tasks such as switching from one activity to another. In some studies (e.g., Dajani & Uddin, 2015; Uddin, 2021), cognitive flexibility is considered one of the primary constructs of Executive Functioning. In these studies, cognitive flexibility is broadly defined as the ability to perform different tasks, shift attention between these tasks and adjust between multiple concepts according to changes in a given situation.

Working memory refers to the cognitive mechanism that stores, retains, manipulates, and processes information to ensure the successful execution of behavioural responses. Working memory is usually confused with short-term memory. Although there is a large overlap between working memory and short-term memory (Hornung et al., 2011; Martínez et al., 2011), working memory theoretically represents the whole framework of processes used for temporary retainment and manipulation of information, whereas short-term memory is only one of the components within this framework. Working memory is critical for cognitive tasks like problem-solving, reasoning, learning and language comprehension. The construct of working memory can be difficult to quantify as it is often confounded in Executive Functioning tasks, and because working memory itself can be measured in different ways. Performance-based tasks that were used to measure cognitive flexibility such as WCST and D-KEFS usually involve working memory abilities to retain and manipulate information during the task (Geurts et al., 2009). It is challenging to separate performances that explicitly demonstrate cognitive flexibility and working memory.

Response inhibition (also known as inhibitory control) refers to the ability to stop an automatic and dominant behavioural response to a given stimuli (Brian et al., 2003). A range of response inhibition tests such as negative priming, Stop-Signal and Go/No-Go tasks to evaluate the inhibition performance of typically developing individuals (Ozonoff & Jensen, 1999; Ozonoff & Strayer, 1997). Response inhibition is also an important construct in Executive Functioning which plays a significant role in the development of Emotion Regulation.

### 1.2.1 Executive Functioning in Typical Development

Executive Functioning manifests around three years of age (Posner & Rothbart, 1998). In Posner and Rothbart's (1998) study, they investigated the development of executive attention using marker task strategy tracing the activation of brain areas from neuroimaging studies. The task required the child to press a button when a visual stimulus was shown on one side of the screen. During the congruent trial, the child was trained to press the button on the side where the stimulus was presented, whereas during the incongruent trial, the child was required to inhibit the predominant response and press the button on the side opposite the stimulus. Executive Functions such as response inhibition, executive attention and cognitive flexibility were involved in this task, and thus, an increase in accuracy indicated development of these abilities. They found strikingly different patterns of performances between typically developing children at the end of three years old and children at the beginning of four years old on a Stroop-like task, and their performances reached high accuracy by the age of 36 to 38 months. These findings support Kochanska et al.'s (2000) longitudinal study which also found significant improvement on Executive Functioning tasks in typically developing children at the age of 22 and 33 months, suggesting that a noteworthy improvement and significant development of Executive Functioning during the age of 22 to 38 months.

During toddlerhood, Executive Functions such as basic response inhibition emerges allowing children to suppress motor responses (Garon et al., 2008) and early forms of working memory to support brief retention and manipulation of information (Diamond, 2013). This period is marked by a gradual increase in the ability to wait and delay gratification. Waiting involves being patient and enduring a period of time without immediate reward. It also involves the ability to inhibit emotional impulses and engage in goal-directed behaviours. Children who can wait demonstrate better emotion regulation (Carlson & Wang, 2007). Delay of gratification refers to the ability to resist the temptation of an immediate reward in favour of a more significant reward that will be available later. Similar to waiting, successful delay gratification involves resisting the emotional pull of immediate rewards and focus on the long-term goals (Mischel et al., 1989).

Advances in response inhibition become evident during preschool stage aiding in the control of impulsive behaviours (Best & Miller, 2010) and decision-making (Luna et al., 2010). Working memory capacity expands during this stage, allowing for more complex cognitive tasks (Gathercole & Pickering, 2000) and early development of cognitive flexibility is observed, allowing for simple task switching (Zelazo et al., 2003). Improved response inhibition supports the regulation of emotional impulses and enhanced working memory enables children to hold and manipulate information relevant to emotion regulation strategies (Carlson et al., 2004). Improvement in cognitive flexibility also helps children adapt emotionally to changing situations and perspectives (Zelazo et al., 2003). Executive Functioning milestones in typical development are closely related to Emotion Regulation and the development of Executive Functioning is intertwined with the maturation of

Emotion Regulation abilities. An overview of Emotion Regulation and its developmental milestones will be provided in the next section. The relationship between Executive Functioning and Emotion Regulation will also be discussed further.

## 1.3. Emotion Regulation (ER)

Emotion Regulation is conceptualised as the ability to modulate one's emotional arousal to foster an optimal level of engagement with the environment (Cicchetti et al., 1991). This ability is critical both in initiating, motivating, and organizing adaptive behaviour, and in preventing stressful levels of negative emotions and maladaptive behaviour. The score of Emotion Regulation in the current research include both Emotion Regulation ability and Emotion liability-negativity. The latter can be described as one's ability in recovering from negative emotion reactions (Dunsmore et al., 2013).

During the first month of life, infants experience changes in their arousal level, leading to physiological discomfort. They learn to attain physiological balance to reduce the internal tensions with motoric reflexes, which gives rise to the development of emotion regulation to modulate their emotional states (Cicchetti et al., 1991). However, not all internal needs can be met with infants' internal regulation; they require help from the environment and rely on their caregivers to provide extrinsic modulation. Infants learn to seek for their caregivers to communicate their needs effectively with gestures or readable expressions (Pollak et al., 2019). Moving from infancy to toddlerhood, children gradually develop more self-sufficient abilities in modulating and regulating their emotional states (Gross, 2014; Kopp & Neufeld, 2003). When they encounter a novel or an unpleasant situation, they start engaging in basic Emotion Regulation strategies such as self-soothing (e.g., finger sucking, body rubbing; Buss & Goldsmith, 1998) and self-distraction behaviours (e.g., moving from one activity to another and looking away; Zimmer-Gembeck & Skinner, 2011) to relieve discomfort.

Emotion reaction is relatively a more automatic response that are not consciously controlled in early childhood, which can result in socially inappropriate behaviours (Barrett & Campos, 1987), such as shouting, screaming and temper tantrums. These emotions could be evoked by internal factors, such as atypical arousal threshold or sensory sensitivity, or external factors, such as changes in routine, incompleteness, demands, or stimulating environments. Between the ages of 2 and 5 years, children develop more sophisticated forms of self-regulation ability in a more socially acceptable manner (Carlson & Wang, 2007). In Aldao and Nolen-Hoeksema's (2012) point of view, successful Emotion Regulation strategies are adaptive, and unsuccessful Emotion Regulation strategies are known as maladaptive Emotion Regulation strategies, in which whether an Emotion Regulation strategy is adaptive or maladaptive is determined by the context of the situation. In general, adaptive Emotion Regulation strategies include behaviours such as seeking help, comfort, social support, restructuring thoughts, and changing the way appraising an aversive situation. Maladaptive Emotion Regulation responses take the form of avoidance, aggression, temper tantrums, repetitive behaviours and self-injurious behaviours (Samson et al., 2014).

### 1.3.1 How Executive Functioning relate to Emotion Regulation in Typical Development

Improvements in children's ability to regulate their emotions and behaviours were evident in line with performance gains in Executive Functioning tasks. For example, Li-Grining's (2007) longitudinal study observed improvement in waiting behaviours in 439 children. These typically developing children who were between 2 to 4 years old, took part in two delay-of-gratification tasks (i.e., Snack delay and Gift Wrap) during the first session and again 16 months later along with two additional executive control tasks (i.e., Shapes task and Turtle-Rabbit task). During the Snack Delay task, an M&M candy was presented to the children, and they were asked to withhold from eating them for a duration of 20 seconds to 60 seconds, until the experimenter rang a bell. During the Gift Wrap task, children were presented with a wrapped gift, and they were asked not to peek at it for 60 seconds. Children were presented the same delayed-gratification tasks 16 months later and improvement in these tasks were observed. Results showed that older children performed significantly better than younger children on both delayed gratification and executive control tasks. However, the study did not include executive control tasks during the first session and did not examine whether the improvement in delayed gratification tasks were correlated with executive control tasks. Nevertheless, children's ability to control their emotional responses improve around the same time as Executive Functioning. According to Fuster et al. (2009), as Executive Functioning becomes more mature, children become more aware of their internal states and can control their emotional reactions. The development of Executive Functioning and Emotion Regulation ability during the same time period leads us to consider that they may be related.

Response inhibition, a construct of Executive Functioning, is established at the age of 4 (Eisenberg et al., 2007) allowing the child to inhibit an automatic response and exhibit another socially appropriate response to achieve goals. Improvement in response inhibition enables children to withhold their responses during situations that trigger automatic emotional reactions, take control in these reactions, and then reorient their responses to adapt social expectations. Carlson and Wang (2007) examined the relationship between response inhibition and Emotion Regulation of 53 typically developing preschool children, aged 4 to 6 years. They administered three tasks to measure children's response inhibition (i.e., Simon Says, Forbidden Toy and Gift Delay) and three tasks to measure their emotion regulation (i.e., Emotion Understanding, Secret Keeping, and Disappointing Gift). In addition to these tasks, children's ability was evaluated using the Peabody Picture Vocabulary Test (3rd ed.; Dunn & Dunn, 1997) and their parents completed two questionnaires: Self-Control Rating Scale and Emotion Regulation Rating Scale to obtain parent-report information regarding the children's ability in regulating their behaviours and their emotions. The study found that children's performances in response inhibition tasks were significantly correlated with their ability to regulate their emotions and this relationship remained significant even after controlling for the effects of age and verbal ability. The parental report of self-control and emotion regulation were also highly correlated. These findings further demonstrate that executive functions are related to emotion regulation.

Schmeichel et al. (2008) examined the relationship between working memory capacity and emotional experiences with a sample of 45 undergraduate students between 18 to 23 years old. The participants first completed the Berkeley Expressivity Questionnaire (BEQ; Gross & John, 1997) and a working memory test (OSPAN task; Turner & Engle, 1989). Then, they were shown film clips that were likely to trigger unpleasant emotions. Schmeichel et al. (2008) reported that a higher level of working memory correlated with a higher ability to modulate the unpleasant emotion following an aversive situation. In another study comprising 89 typically developing participants between the ages of 18 to 36 years old, McRae et al. (2012) also found a moderate positive correlation between adaptive emotion regulation strategy (cognitive reappraisal) and Executive Functioning (working memory and cognitive flexibility). This finding suggests that the ability to hold and process information is likely to relate to the ability to regulate emotion. These results were further strengthened by Malooly et al. (2013), who found that greater cognitive flexibility was related to a higher ability to reduce the experience of sadness following sad film clips. These findings indicate the potential relationship between Executive Functioning and Emotion Regulation.

A causal relationship between these two constructs was suggested by Li et al.'s (2020) study when they found executive function training improved children's emotional competence. Fifty-five four-year old children were recruited and assigned into two groups: training group ($n$ = 29) and no-training group ($n$ = 26). The training took place twice a week over two months with 12 sessions in total. The first four sessions aimed to develop response inhibition ability, the fifth and sixth sessions aimed to promote cognitive flexibility, the seventh and eighth aimed to foster working memory, the ninth and tenth aimed to improve problem-solving ability and the last two sessions were used to review all the training sessions. The emotional competence and Executive Functioning of children from both groups were assessed before and after the training. Emotional competence was assessed with 1) Facial Expression Match and Recognition, 2) Test of Emotion Comprehension (TEC), 3) Situational Storytelling of Emotion Expression, and 4) Situational Storytelling of Emotion Regulation. Their executive functions were measured with 1) Emotional Stroop Test (response inhibition), 2) Dimensional Change Card Sorting (cognitive flexibility), 3) Memory for Picture of Wechsler Intelligence Scale for Children (IV; working memory) and 4) Situational Storytelling of Problem Solving (problem solving). Results revealed that children from the training group scored significantly higher on emotional competence tasks during post-test than no-training group, but not during pre-test, suggesting that children's emotion competence improved following the Executive Functioning training. The authors also examined the difference in performance on Executive Functioning tasks and emotion competence tasks between pre-test and post-test. They found that improvement on

Executive Functioning tasks was positively correlated to the improvement on emotion competence tasks ($r = 0.49$, $p < 0.01$). From this study, we know that Executive Functioning was not only correlated with the ability to regulate emotion, but that changes in Executive Functioning predicted the change in Emotion Regulation.

Compared to typically developing individuals, individuals with autism employ more maladaptive Emotion Regulation strategies and fewer Emotion Regulation adaptive strategies (Cai et al., 2019; Jahromi, 2017; Samson et al., 2015). One of the hallmark characteristics in autism, Restricted and Repetitive Behaviours (RRBs), potentially serve as a strategy to regulate emotions and sensory experiences (Mazefsky et al., 2013). While South et al. (2007) was investigating the relationship between Executive Functioning and RRBs in autism, they also found a complex relationship between RRBs and Emotion Regulation, emphasizing the need for a nuanced understanding of how these behaviours may function in different context.

## 1.4. Restricted and Repetitive Behaviour (RRBs) in Typical Development

Restricted and Repetitive Behaviours (RRBs) refer to behaviours ranging from repetitive body movements (e.g., hand-flapping, body rocking, repetitive vocalisation) to restricted interests and routine (e.g., attaching to a particular type of foods or toys). These behaviours can be observed throughout typical development (Arnott et al., 2010; Harrop et al., 2014; Leekam et al., 2007; Uljarević et al., 2017). Thelen's (1979) developmental account posited that repetitive motor movements play an essential role in neuromuscular and motor development. Children start exhibiting repetitive behaviours in their first year of life, and around two years old, at its peak, they spend almost forty per cent of their time engaging in these behaviours (Thelen, 1979).

Toddlers display repetitive motor movements in typical development to develop necessary postural control for a new body position or a complex motor movement (Thelen, 1980). For example, rhythmical kicking movements can be observed around one month of age before the infant can move their legs for crawling and supported stepping; finger flickering precedes the ability of goal-directed grasping. Hand-flapping and arm-waving allow toddlers to explore the mobility of their arms through repetitive tuning of the timing and amplitude of muscle activation, ultimately facilitating more precise and efficient reaches (Thelen, 1979; Thelen & Cooke, 1987). Shafer et al. (2017) supported this framework by claiming that the motor system restricts the degree of freedom of motor movements in the early state of motor learning, to gain control and produce simple and stable movements, such as repetitive kicking and hand swaying. Following the maturation of cortico-striatal circuits and the motor system and when learning progresses, usually at the end of the first year of normative development, the system expands the degree of freedom to allow more specific and more accurate motor movements (Shafer et al., 2017). Toddlers have more control over their physical movements, and their motor movements become increasingly goal-directed (Leekam et al., 2011).

RRBs also serve the purposes of adaptive functioning and play a transitory role in normative development. During the first year of life, typically developing infants have little understanding of the contingency of their behaviours, others' behaviours, and the environment. They do not have much control over their environment as they have limited adaptive behaviours, such as functional communication to avoid undesired edibles, activities, and objects. A range of RRBs can be observed during this period, such as ritualistic behaviours (e.g., attaching to a particular object; Zohar & Felz, 2001), rhythmical stereotyped behaviours (e.g., being carried and bounced by caregivers; Thelen, 1979), and repetitive motor movements (e.g., swaying and body rocking; Leekam et al., 2007). Zohar & Felz (2001) examined the relationship between ritualistic behaviours, fearfulness and emotional temperament in typically developing children below 6 years. Mothers rated the child's temperament, fearfulness, and ritualistic behaviours. Results showed that children with more ritualistic behaviours had higher level of fearfulness, temperament dimensions of negative emotion, and shyness. Zohar & Felz (2001) suggested that this result was consistent with the developmental theory which claimed that ritualistic behaviours, during the early age, serve as adaptive behaviours by introducing order and predictability in their environment and consequently, reduce their worries, fear and other negative emotions (Evans et al., 1997; Zohar & Felz, 2001b). Therefore, RRBs have also been considered a type of arousal and Emotion Regulation behaviours during early childhood (Leekam et al., 2011; Uljarević et al., 2017; Zohar & Felz, 2001).

In addition to the relationship between Emotion Regulation and RRBs, Executive Functioning is also closely related to RRBs. RRBs decline over time and reduce significantly after four years of age (Çevikaslan et al., 2014; Leekam et al., 2011; MacDonald et al., 2007; Uljarević et al., 2017) when children begin to advance in cognitive skills and goal-directed actions (Leekam et al., 2011), which also coincides with the manifestation of Executive Functioning, a set of mental processes that are involved in the cognitive control of behaviour. However, children at risk for and identified with Autism Spectrum Conditions (ASC) and developmental disabilities start exhibiting repetitive behaviours later than typically developing children (Symons et al., 2005), and these behaviours can still be observed at high frequency after four years old (Bodfish et al., 2000; Richler et al., 2010; Wolff et al., 2014). Deficits in Executive Functioning have often been observed in individuals with ASC, which eventually led to the formation of the executive dysfunction hypothesis

(Demetriou et al., 2018). The synchronicity of the decline of RRBs and manifestation of Executive Functioning, together with the executive dysfunction in ASC which are likely to relate to the maintenance of RRBs at high frequency after toddlerhood, suggest a potential relationship between RRBs and Executive Functioning.

The above studies indicate a possible relationship between Executive Functioning, Emotion Regulation and RRBs in typical development and potentially, in autism. In the next section, we will explore these three constructs in autistic population.

## 1.5. Autism Spectrum Condition (ASC)

Autism spectrum condition (ASC) is a neurodevelopmental disorder typically characterised by challenges with social interactions, speech and nonverbal communication, restricted interest, repetitive motor movements and preoccupation with objects (American Psychiatric Association, 2013), that occurs in various forms and on a broad continuum of severity. Some individuals with ASC experience mild challenges in everyday life, while others experience a more significant impact in their life. The awareness regarding ASC in Malaysia is gradually increasing but still lower compared to neighbouring countries such as Singapore (Ilias et al., 2017). There is no local official epidemiological data available on the prevalence of ASC in Malaysia but the World Population Review (2022) identified approximately 81.60 per 10,000 or 1 in 122 children in Malaysia as having ASC, and the number rises by three per cent every year (Zakaria, 2016).

In general, clinicians are still widely divided on whether this condition can be reliably identified and diagnosed before three years old (Stone et al., 1999), even though most of the symptoms of ASC manifest during infancy. Ninety per cent of the parents noticed the first symptoms of ASC when the children were approximately 19 months old (De Giacomo & Fombonne, 1998). Failure to diagnose ASC at an early age is likely to hinder the implementation of early interventions (Ilias et al., 2017). Still, the process can be very challenging due to the wide variety of autistic characteristics, which affect the reliability and stability of diagnosis across clinicians (Pierce et al., 2019). Understanding the progression of the diagnostic classification system for ASC could help us identify the challenges both clinicians and researchers face.

Autism was first introduced in DSM-III, but Asperger Syndrome gained more public appreciation when Lorna Wing introduced the concept of “spectrum” for autistic disorders (Wing, 1991). This conceptualization led to the inclusion of Asperger Syndrome in the Diagnostic and Statistical Manual of Mental Disorder ($4^{th}$ rev.; DSM-IV; American Psychiatric Association, 1994) and the International Classification of Diseases (10th rev.; ICD– 10; World Health Organisation, 2004). In the DSM-IV, the term “Pervasive Developmental Disorders (PDD)” was used as an umbrella term encompassing five sub-disorders, all characterised by deficits in social interaction, and communication, restricted interests, and repetitive movements. The five disorders categorised under PDD were 1) Autistic Disorder, 2) Asperger’s Disorder, 3) Pervasive Developmental Disorder, Not Otherwise Specified (PDD-NOS), 4) Rett’s Syndrome, and 5) Childhood Disintegrative Disorder (CDD; American Psychiatric Association, 1994). Due to the lack of consistency among clinicians in assigning the diagnostic subtypes, the American Psychiatric Association (APA) updated this classification system in 2013. They replaced the first three disorders with a single term “Autism Spectrum Disorder (ASD),” whereas Rett’s Syndrome and CDD no longer fell under the ASD diagnosis. Nowadays, most clinicians refer to the Diagnostic and Statistical Manual of Mental Disorder ($5^{th}$ rev.; American Psychiatric Association, 2013) for diagnostic criteria for ASC.

Aside from the changes in the diagnostic subtypes, the DSM-5 also combined both language communication and social interaction deficits into the same category, considering that communication is inevitably social in nature. Rather than having three diagnostic criteria, the DSM-5 has only two main diagnostic criteria for ASC: 1) persistent deficits in social communication and social interaction and 2) restricted, repetitive patterns of behaviour, interests or activities. The latter features of ASC contained four subcategories in DSM-IV: (1) repetitive and stereotypical motor movement, (2) ritualised patterns of verbal or non-verbal behaviours, (3) highly restricted interest, and (4) preoccupation with objects. The DSM-5 maintained the first three categories and replaced the last one with "hyper-or hypo-sensitive to sensory aspects of the environment." The field trial for ASC in the DSM-IV examined these sensory features but did not find them powerful diagnostic features for ASC (Grapel et al., 2015). Studies later reported that 66% to 90% of individuals with ASC have atypical sensory interests and reactivities (Leekam et al., 2007; Zachor & Ben-Itzchak, 2014), which led to the changes in the DSM-5, but the rationale of the inclusion has been questioned due to the lack of epidemiological studies. It is important to note that many studies have examined the relationship between sensory reactivity in ASC and repetitive behaviours and claimed that sensory reactivity predicts repetitive behaviours in autistic children (Gabriels et al., 2008; Kirby et al., 2017; Schulz & Stevenson, 2019). The inclusion of atypical sensory reactivity in the DSM-5 under the predominant diagnostic criteria of RRBs suggests that hyper- and hypo-reactivity can play an important role in these behaviours. It should be noted that sensory reactivity differences are not unique to ASC, and are found in conditions such as Sensory Processing Disorder (SPD) and Attention-Deficit/Hyperactivity Disorder (ADHD).

## 1.6. RRBs in ASC

In comparison to social and communication deficits, there is a noticeable lack of research on restricted and repetitive behaviours (RRBs), perhaps due to their inconsistent conceptualisation and the lack of standardised measurements. RRBs contain a broad range of characteristics that may appear in combination among individuals with a different severity in ASC. Some RRBs such as circumscribed interests can be considered a strength in careers that involve specific expertise such as advanced skills in computer science (Leekam et al., 2011). However, these behaviours are regularly rated by caregivers of children with ASC as being the most challenging features of ASC to manage (Bishop et al., 2007) and often lead to higher stress levels for parents and family members (Lecavalier et al., 2006). RRBs have also been found to affect the quality of family well-being (Lounds et al., 2007) and engender negative parenting styles (Greenberg et al., 2004). Besides these negative impacts on caregivers and families, these behaviours can dominate the daily life of individuals with ASC (Harrop et al., 2016), and constitute major barriers to the development of functional skills (Cuccaro et al., 2003), learning opportunities (Pierce & Courchesne, 2001), and social engagement (Loftin et al., 2008; Nadig et al., 2010).

Research in RRBs is heavily affected by the conceptualisation and description of these behaviours and any changes in their definition especially in its diagnostic criteria can have a noteworthy impact on autism research. A recent meta-analysis by Iversen & Lewis (2021) noted a rapid increase in research on RRBs following the definitional changes in the DSM-5. They searched for articles related to RRBs in the ISI (Clarivate Analytics) Search Engine and found only 258 articles between 2008 and 2013, compared to 767 articles between 2014 and April 2020. The conceptualisation and description of RRBs have been studied and discussed over the past 20 years. Although there is a broad agreement that

RRBs are generally related to repetitive movements, rigidity and restricted interest, there are continuing arguments whether RRBs should be conceptualised as a uni-dimensional or multi-dimensional model with several relevant but distinct features and behavioural categories.

### 1.6.1. Factor analytic studies of the RRBs domains

Early studies (e.g., Prior & Macmillan, 1973; Turner, 1999) conceptualised RRBs into dichotomous groups, “lower-order” and “higher-order” behaviours, based on clinical observations. The “lower-order” RRBs, also known as sensory-motor repetitive behaviours, referred to repetitive motor movements such as arm swaying, finger flickering, and a preoccupation with objects, whereas “higher-order” RRBs referred to restricted interests, ritualised behaviours, and routine. “Lower-order” RRBs have been observed more frequently in developmentally delayed children with a lower level of cognitive and language abilities, and “higher-order” behaviours have generally been observed in children with a higher level of cognitive and language abilities (Bishop et al., 2006; Esbensen et al., 2009). A study comprising a sample of 192 parents of typically developing children with no diagnosed medical conditions, reported that the “lower-order” RRBs and the “higher-order” RRBs develop independently from each other from 15 to 77 months (Uljarević et al., 2017).

This dichotomous framework was later supported by the findings from a principal component analysis (PCA; Cuccaro et al., 2003) using the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994). The ADI-R is one of the most widely used instruments in autism research in determining whether or not individuals have ASC. This 93-item semi-structured interview was administered to parents or caregivers of 292 individuals with ASC between the ages of 3 to 21 and a two-factor solution provided a good fit for the data, accounting for 32% of the variance. In another EFA (Lam et al., 2008), only 10 items of the

ADI-R were administered, and three-factor solution were found: “Repetitive Motor Behaviours (RMB)”, “Insistence on Sameness (IS)”, and “Circumscribed Interests (CI)”. Sensory items were excluded as they were not part of the diagnostic criteria of ASC until the DSM-5 was published. On the other hand, the Repetitive Behaviour Scale-Revised (RBS-R, Bodfish et al., 2000), which was created specifically for evaluating RRBs, found a five-factor solution: Stereotypic, Self-injurious, Compulsive, Ritualistic/ Sameness, and Restricted Behaviours.

The classification of RRBs is highly varied across studies regardless of the instruments used. The reviewed studies seem to consistently demonstrate the presence of sensory motor repetitive behaviours factor that resemble the “lower-order” RRBs proposed by Turner (1999). The group of behaviours that Turner (1999) termed as “higher-order” RRBs are more varied and less consistent. The factors that can be categorised under the “higher-order” RRBs range from compulsive behaviour, resistance to change, insistence on sameness, ritualistic behaviour, to restricted interest. In general, findings from the reviewed studies seem to lend support to the categories listed under the current diagnostic criteria of RRBs, which are repetitive speech and motor movements, resistance to change, restricted interest and unusual sensory sensitivity.

## 1.7. Executive Dysfunction in ASC

Scientists have been explaining the challenges observed across the life span in ASC including the occurrences of RRBs with a number of cognitive accounts, such as theory of mind (Baron-Cohen et al., 1985) and central coherence (Happé & Frith, 2006b). Executive dysfunction hypothesis is one of the main three cognitive models to emphasise a core role of deficits in Executive Functioning in ASC (Demetriou et al., 2019; Hill, 2004; Lee et al., 2021a). This theory uses deficits in Executive Functioning such as difficulty in initiating new non-routine actions and the tendency of being stuck in an on-going activity to explain behaviour problems in autism such as rigidity and perseveration.

Leung and Zakzanis (2014) examined 72 studies, which was 43 more studies than those gathered by Geurts et al. (2009), including a total of 2,137 individuals with ASC and 2,185 typically developing individuals range 5 to 64 years, using standard meta-analytic statistical techniques. They indicated that lifespan Executive Functioning impairment could be observed in ASC, and this impairment is widely evident in their behaviours. Kouklari, Tsermentseli, and Auyeung (2018) measured the Executive Functioning of 33 participants with ASC and 32 typically developing participants between 8 to 12 years old and found participants with ASC performed significantly poorer in tasks devoted for measuring cognitive flexibility (the Delis-Kaplan Sorting Test) and response inhibition (the Delis-Kaplan Word/Colour Interference) but not working memory (the Delis-Kaplan Word/Colour Interference). Golshan et al. (2019) further supported the presence of cognitive flexibility deficits in ASC with three subtests from NEPSY-II, a neuropsychological assessment measuring Executive Functioning domains (Korkman et al., 2007), and the CHEXI questionnaire (Childhood Executive Functioning Inventory; Catale et al., 2015). These studies serve as an important reminder that the selection of performance-based tasks and rating measures should be carried out with caution because they assess different underlying constructs of Executive Functioning which may subsequently affect the results of general Executive Functioning performance. The measurement of Executive Functioning will be discussed in detail in Chapter 3.

### 1.7.1. Executive Dysfunction and RRBs

A recent spate of research findings lends renewed support to the correlation between heightened RRB levels and executive dysfunction such as deficits in cognitive flexibility (Jones et al., 2018; Miller et al., 2015) and response inhibition (Jones et al., 2018; Mosconi et al., 2009; Thakkar et al., 2008). Miller et al. (2015) examined 60 adults with ASC with a test of cognitive flexibility and found that individuals with difficulty shifting and maintaining new responses, which indicates poorer cognitive flexibility, has heightened RRBs level. Similarly, Jones et al.'s (2018) study revealed notable relationships between RRBs and Executive functioning constructs such as cognitive flexibility as well as response inhibition, but not planning, when they examined 100 adolescents with ASC using 10 tasks to measure the domains of Executive Functioning and theory of mind. As mentioned previously, cognitive flexibility is an ability to shift from ongoing responses to novel responses whereas response inhibition is an ability to inhibit any ongoing responses. These theories of executive dysfunction in ASC posits that deficits in these Executive Functioning constructs lead to individuals with autism becoming "locked in" to a particular set of cognitions or behaviours, such as RRBs (Demetriou et al., 2018).

The relationships between impairment in Executive Functioning constructs and RRBs are not pervasive. Ozonoff et al. (2004) failed to find a relationship between RRBs and cognitive flexibility when examined 79 individuals with ASC and 70 typically developing individuals with Cambridge Neuropsychological Test Automated Battery (CANTAB) subtests across the age range of 6 to 47 years. Additionally, the relationship between RRBs and response inhibition was not significant in Joseph and Tager-Flusberg's (2004) study when they examined 31 children with ASC with five tasks such as Word Span, Block Span, Day-Night, NEPSY Knock-Tap and NEPSY Tower. These inconsistent findings of relationship between executive dysfunction and RRBs suggest other potential variables that may be mediating this relationship. Given the huge amount of studies revealing a significant relationship between Emotion Regulation and RRBs, in this thesis, we propose that Emotion

Regulation may be a potential factor mediating the relationship between Executive Functioning and RRBs.

## 1.8. Emotion Dysregulation in ASC

Emotion Regulation has been less studied in the autistic population because this feature has not been seen as a defining characteristic of ASD compared to social communication deficits and RRBs. Mazefsky et al. (2013) conducted a PsycInfo search of peer-reviewed articles in 2012 with the search terms “emotion regulation” and “autism” which produced only 15 articles. In this thesis, we used the same search strategy on October 31, 2020, and 170 articles were found. The growing number of studies shows increasing interest in Emotion Regulation in the autistic population, which is perhaps due to the development of the theoretical framework in the domain of Emotion Regulation in typical development (see Gross, 2014). Emotion dysregulation in children with ASC (Ho et al., 2012; Kirst et al., 2021; Mills et al., 2022; Nuske et al., 2018) and adolescents with ASC (Mazefsky et al., 2014; Shaffer et al., 2022; Sorter et al., 2022) become more prominent following the development of Emotion Regulation theoretical framework. Costescu et al. (2016) examined Emotion Regulation in children with and without ASC using a false feedback technique, a mood induction task (Brenner, 2000), in which children were given feedback about whether they succeeded or failed during a task to induce an emotion. A sample size of 41 children with ASC and 40 control children between 5 and 11 years old were recruited. These children were presented with several pictures with a piece missing, and they had to choose one picture from three options to complete the picture. Before beginning the predetermined frustrating situation, children took part in five training trials in which they received correct feedback on their performance. They received negative feedback regardless of their performance for ten trials during the frustrating situation. During the positive feedback, children were expected to experience positive emotions whereas they were expected to experience negative emotion during the negative feedback. To increase the intensity of their negative emotions, these children were told that they could choose one of the three available prizes in the beginning regardless of their performance. This rule was then changed, and they would only get the prize following positive feedback. Their emotional expressions were coded based on their facial expression, postures and vocalization during the task. They were also told to rate their emotions at the end of the task. The results showed that relative to typically developing children, children with ASC experienced more dysfunctional negative emotions and displayed more maladaptive behaviours such as staring into space, shifting gaze, using inappropriate words, requesting the experimenter do something with a loud voice and throwing objects. The researchers also found an interesting result in which children with ASC employed the same strategy (i.e., reasoning) during the whole task, whereas typically developing children tried alternative strategies when they noticed the initial strategy led to negative feedback. This finding suggested that children with ASC tend to be more rigid than typically developing children when solving problems, which may contribute to emotion dysregulation and then maladaptive behaviours. As described in the previous section, rigidity is related to Executive Functioning skills, particularly cognitive flexibility.

### 1.8.1. Atypical Sensitivity in ASC and Emotions

Sensory modulation difficulties are also well documented in ASC (Hutt et al., 1964). Sensory hyperreactivity is correlated with autistic traits generally in both adults with ASC and typical development (Tavassoli et al., 2014). According to the latest diagnostic criteria, individuals with ASC are sensory seeking, hyposensitive or hypersensitive to sensory information. Individuals who are sensory seeking crave and are fascinated with certain sensory stimuli. They often display behaviours such as staring intently at light or moving objects, sniffing objects or rubbing against walls or furniture to engage in specific sensory experiences. These behaviours may be a stimulatory or regulatory strategy associated with RRBs (Lidstone et al., 2014). Individuals with hyporeactivity means their senses take in too little sensory information and therefore, have difficulties recognising sensation, and might actively seek for sensory input from the environment when they are under-aroused. They may make loud noises, rock back and forth, and look at certain vibrant colours and bright lights for a long period of time, and these repetitive motor movements provide additional stimulation in the state of under-arousal caused by lack of stimulation in the environment (Goodall & Corbett, 1982). The senses of individuals with hyperreactivity take in too much sensory information and may be overly aroused in certain environments. Certain sensory experiences such as sound, touch, smell, taste, visual and proprioception can be overwhelming for them (Tavassoli et al., 2014). Therefore, they may avoid these sensory experiences by getting away from the sensory stimulation, covering their ears to avoid unpredictable or loud sounds, wearing only certain types of clothing, and avoiding certain foods due to their texture. These extreme sensory experiences have often been conditioned as aversive and have often been related to unpleasant feelings such as anxiety.

We know that children with ASC have a higher risk of developing anxiety compared to the prevalence of 2.2% to 27% among typically developing children (Costello et al., 2005; Gotham et al., 2013; van Steensel et al., 2011; Vasa & Mazurek, 2015)。 In a meta-analysis of 31 studies involving 2121 individuals with ASC below 18 years old, van Steensel et al. (2011) reported that approximately 40% of young individuals with ASC displayed anxiety symptoms across autism studies. A more recent meta-analysis of 81 studies by van Steensel and Heeman (2017) further confirmed youths with ASC have significantly higher levels of anxiety than typically developing youths. Anxiety symptoms persist from childhood to adolescence to adulthood (Ranøyen et al., 2018). Studies have continuously established that although anxiety is not considered a core feature of ASC, many individuals with ASC experience high anxiety. Diagnostic criterion of autism such as hyperreactivity, hyporeactivity and sensory seeking have been related to anxiety in ASC (Green et al., 2012; MacLennan et al., 2021, 2022). A study involving 54 children with ASC, age 3 to 5 years found anxiety correlated with sensory hyperreactivity, in which intolerance of uncertainty served as an important interrelated construct (MacLennan et al., 2021). Verhulst et al. (2022) asked autistic adults regarding the sequence of sensory and anxiety and they found that hyperreactivity precedes anxiety whereas anxiety precedes sensory seeking.

Autistic individuals especially those with hyperreactivity are highly sensitive to sensory information from the environment but sensory modulation deficits can limit their ability to filter and regulate the overwhelming inputs. These deficits make their world extremely unpredictable and are more likely to interpret novel sensory stimuli as threatening or dangerous. Social communication deficits further increase the challenges of regulating their sensory experiences and emotions because they can limit the ability of autistic individuals to express their emotions, seek for comfort and communicate to remove the environmental stimuli that cause distress. There is also an increasing number of studies relating the manifestation of anxiety to impaired Emotion Regulation (Cai et al., 2018; Conner et al., 2020; White et al., 2014), which is common in ASC (Hill, 2004; Mills et al., 2022; Samson et al., 2014). In summary, sensory sensitivity has been related to anxiety, which potentially relate to Emotion Regulation, which has been linked to RRBs.

### 1.8.2. RRBs as an Emotion Regulatory Mechanism

There has been increasing interest in the role of repetitive behaviours as an emotional regulatory mechanism for autism. Some studies have suggested that individuals with ASC employ RRBs (e.g., sensory motor repetitive behaviours) to seek sensory input when they are under-aroused to reduce anxiety (Joosten et al., 2012; Lovaas et al., 1987). Other studies have suggested that these behaviours help avoid certain sensory inputs and reduce anxiety (Joosten & Bundy, 2010). In a study that involved 54 parents of typically developing children and 50 parents of children with ASC, Black et al. (2017) found that sensory hyperreactivity mediated 67% of the relationship between anxiety symptoms (specific phobia) and RRBs (insistence on sameness), and 57% of the relationship between separation anxiety and insistence of sameness. Lidstone et al.'s (2014) study further supported this result finding that RRBs, particularly insistence on sameness, reduced anxiety through avoidance of sensory stimulation. In this study, 120 parents of 2- to 17-year-olds with ASC completed the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007) and 49 of them later completed the Spence Anxiety Scales and the Sensory Profile. Results showed that anxiety predicted insistence on sameness and sensory avoidance. Sensory avoidance predicted insistence on sameness but the relationship between insistence on sameness and anxiety was no longer significant when sensory avoidance was included. This result implies that children with ASC enforced ritualistic and rigid behaviours to avoid overwhelming sensory input, which in turn reduced anxiety.

The role of repetitive behaviours serving as a coping mechanism to reduce anxiety was first mentioned in Kanner's (1943) most cited paper, "The child's behaviour is governed by an anxiously obsessive desire for the maintenance of sameness that nobody but the child himself may disrupt on rare occasions. Changes of routine, of furniture arrangement, of a pattern, of the order in which daily acts are carried out, can drive him to despair." (p. 245). Changes in the daily routine can lead to distress for the child. Kanner (1943) continued, "The dread of change and incompleteness seems to be a major factor in the explanation of the monotonous repetitiousness and the resulting limitations in the variety of spontaneous activity" (p. 246). Kanner described the need for sameness and repetition to provide a state of calmness and to maintain a body and mind in homeostasis. Kanner's descriptions significantly influenced the development of theoretical frameworks for repetitive behaviours in autism. Anxiety is one of the most heavily studied emotions in the research of RRBs (Joyce et al., 2017; Lidstone et al., 2014; Muskett et al., 2019; Rodgers et al., 2012; K. M. Russell et al., 2019). These studies found high levels of anxiety were associated with high levels of RRBs and vice versa, suggesting that RRBs are mechanisms to cope with anxiety and to regulate emotions.

Most studies in the field have only found anxiety related to high-order RRBs but not lower-order RRBs. In a sample of 67 children with ASC between 8 and 16 years old, children with high anxiety had more repetitive behaviours than those with low anxiety (Rodgers et al., 2012). In the high anxious sample, higher-order RRBs, particularly insistence on sameness, were associated with higher level of anxiety symptoms. Lower-order RRBs (sensory motor repetitive behaviours) were not associated with anxiety level in this sample. Interestingly, one anxiety subtype (i.e., Obsessive-Compulsive Disorder, OCD) was associated significantly with sensorimotor repetitive behaviours in the low anxious sample. The authors explained that this result was perhaps due to the overlapping features between OCD and sensory motor repetitive behaviours which can be difficult to differentiate. However, this study failed to consider the children's sensory sensitivity and Emotion Regulation ability of the children from both anxious and non-anxious group. As discussed in the previous section, individuals might engage in sensory motor repetitive behaviours as an outcome of anxiety (Joosten et al., 2012) and might use rigidity and ritualistic behaviours to avoid sensory stimulation to reduce anxiety (Black et al., 2017; Lidstone et al., 2014). Taking sensory stimulation and emotion regulation ability into consideration can facilitate understanding of the different types of RRBs used in different contexts.

Higher-order RRBs such as insistence on sameness and ritualized behaviours were also related with other emotions such as depression (Stratis & Lecavalier, 2013). RRBs have also been observed in fearful situations. Hirschler-Guttenberg et al. (2015) examined Emotion Regulation strategies of 40 children with ASC between 3 to 7 years old and 40 typically developing children between 2.5 to 7 years old at the child's home. The children's behaviours were observed in three situations: neutral, fearful and positive. During the neutral situation, the parent and child engaged in free play with a predetermined toy for 7 minutes. The fearful situation involved the parent and the child sitting in front of the experimenter, who put on four masks likely to elicit increasing fear: rabbit, lion, alligator, and monster, for 15 seconds each. Lastly, the parent and the child played with colourful hand puppets together for 5 minutes in the positive situation. Children with ASC displayed less positive emotionality than control children across these three situations indicating that reduced positive emotional expression is not a consequence of an aversive situation. Compared to control children, children with ASC used simpler Emotion Regulation strategies such as repetitive self-talk, physical self-soothing behaviours (e.g., repetitive thumb-sucking), idiosyncratic behaviours (e.g., hand flapping), venting and avoiding, to manage their emotions, particularly during fearful situations.

RRBs also serve as an outlet for excitement. In Willemsen-Swinkels et al.'s (1998) study, 54 children with developmental problems (e.g., autism, attention deficit/hyperactivity disorder, developmental language disorder), and 28 children with typical development were recruited. The behaviours of these children were observed while interacting with one of their parents. Of these 82 children, 26 children showed sensorimotor behaviours. Their age ranged from 33 to 84 months old and of these 26 children, 14 children had a PDD, two children had an attention-deficit/hyperactivity disorder (ADHD), six children had a developmental language disorder, and four children did not match any DSM-IV diagnosis and were thus considered typically developing. The children’s behaviours were videotaped during a 40-minute semi-structured playroom session. They were separated from their parents for 3 minutes and reunited later. Following the reunion, the parents read silently and performed three tasks: blowing bubbles, watching television, and building a wall with blocks with their children. Finally, there was a period of free play. During the session, the duration of the repetitive behaviours was recorded. The children's emotion was judged whether they were experiencing elation, distress or composure based on their facial expressions and the content of verbal or nonverbal communication. Their heart rate was recorded throughout the session to explore whether the rate changes when the children were presented with different emotional contexts. The results found that children’s heart rates peaked right before the onset of the repetitive behaviours across different situations. Repetitive behaviours could be an effective coping strategy that served a calming function when children were experiencing distress. The behaviours compensated for an overstimulating environment, helped the child switch attention away from the environment, and calmed down. The reduced heart rate following the repetitive behaviours suggested decreased arousal because these repetitive behaviours soothed the external stimulation. Unlike the distressed situation, when the child was excited (elation situation), the repetitive behaviours served as an expression of excitement without redirecting the child’s attention.

Repetitive behaviours during calm situations were not associated with heart rate, which led the author to posit that these behaviours might serve social functions that were different from elation and distress situations. This study has also found that children with IQs below 50 used repetitive behaviours frequently during all three conditions (distress, elation and composure), children with moderate IQs displayed these behaviours in elation and composure conditions, whereas children with IQs above 80 displayed these behaviours only during the composure condition. These findings imply that cognitive abilities might affect the number of repetitive behaviours in different emotional contexts.

In conclusion, many past studies have shown a relationship between Emotion Regulation and RRBs and supported the role of RRBs as an Emotion Regulatory mechanism across emotional contexts such as anxiety, fear and excitement. The inconsistent findings of the relationship between Executive Functioning and RRBs lead us to consider other potential variables which may be mediating their relationship. Considering the extensive research demonstrating a connection between Emotion Regulation and RRBs, we believe it is important to examine whether Emotion Regulation mediates the relationship between Executive Functioning and RRBs, which is the main goal in this thesis.

## 1.9. Neurobiological Accounts

A neurobiological account of RRBs further posited potential relationships with Executive Functioning and emotions based on the three “macro circuits” theoretical framework (Yerys, 2015a). From a neurobiological perspective, Lewis and Bodfish (1998) argued that RRBs should be viewed in a unidimensional model. These behaviours can sometimes be observed within individuals and across various disorders, despite the distinctive features in ASC. The co-occurrences of RRBs suggest common pathophysiology shared among these behaviours. Lewis and Bodfish (1998) reported that RRBs could be mediated by manipulating the nigrostriatal dopaminergic pathway. However, this unidimensional model was challenged when more refined neurobiological studies identified that different subtypes of RRBs are related to different brain areas. Briefly, the motor and premotor cortex are related mainly to repetitive motor movements, the dorsolateral prefrontal cortex is related mainly to rigidity and ritualised behaviours, and the anterior cingulate and lateral orbitofrontal cortex have been found active during compulsive behaviours (Langen et al., 2011). These findings prompted the proposal of three "macro-circuits" and led to the conceptualisation of RRBs as a multidimensional model. The neurobiological theories and evidence provide another approach to classifying RRBs and further insights into the origin of RRBs in ASC.

Neuroimaging studies focusing specifically on RRBs in ASC are scarce. At first, repetitive behaviour research was mainly led by animal studies and limited to repetitive motor movements, which are also known as motor stereotypies (Lewis et al., 2007). The research direction in this area later transitioned to human disorders following the advancement of neuroimaging and eventually extended to human emotional and cognitive domains. The basal ganglia have been considered a vital brain area for explaining repetitive behaviours. They are a group of subcortical brain structures, including the striatum, globus pallidus, substantia nigra and subthalamic nucleus (Lanciego et al., 2012). Early studies have found correlations between the volumes of the basal ganglia and repetitive behaviours (Garner, 2005; Langen et al., 2011; Norman & Shallice, 1986; Turner, 1997) but results are somewhat ambiguous.

Early studies focused mainly on the role of basal ganglia as a candidate providing a pathway from the cortical areas to the motor cortex and how this pathway affects human repetitive motor movements. The investigation of the role of the basal ganglia expanded from primarily motor-based to cognitive and emotional domains in recent studies (Jahanshahi et al., 2015). A tripartite model was proposed to explain the repetitive behaviours in ASC, suggesting that three networks connecting the cortical regions to the striatal regions may play a role in these behaviours. These networks were usually referred to as cortico-striatal circuits. Researchers hypothesized that any disruptions to these brain networks can result in repetitive behaviours (Langen et al., 2011; Lewis et al., 2007; Lewis & Kim, 2009). The introduction of the tripartite model excited the field because it provided a comprehensive, sensible, and convincing framework which translated directly from animal models. This model has also given rise to the investigation of the potential roles of cognitive and emotion domains in repetitive behaviours.

The development of cortico-striatal circuits framework began when Alexander et al. (1986) argued that the basal ganglia provide multiple parallel and segregated pathways connecting both cortical associative areas and primary motor cortex, while strongly interconnected with other brain areas, such as the cerebral cortex (e.g., premotor and prefrontal cortex), thalamus, and the brainstem. They receive and integrate wide-ranging inputs from these brain areas, supporting sensorimotor, cognitive, and limbic functions. Alexander et al. (1986) proposed five parallel cortico-striatal circuits: 1. the motor circuit, 2. the oculomotor circuit, 3. the dorsolateral prefrontal circuit, 4. the lateral orbitofrontal circuit, and 5. the anterior cingulate circuit. These circuits are closed networks connecting specific cortical areas and striatum, and they were named after the cortical areas. Based on the later functional and structural neuroimaging data, this model was refined, and the cortico-striatal circuits were functionally categorised into three macro circuits: the sensorimotor circuit (movements), the associative circuit (cognitive functions), and the limbic circuit (emotional-motivational behaviours (Groenewegen et al., 2003; Jahanshahi et al., 2015). The types of repetitive behaviour displayed are strongly related to the location of the interruption within the circuit (Mason, 2006).

### 1.9.1. Sensorimotor Circuit

The sensorimotor circuit consists of the sensorimotor and premotor cortex, projecting to the dorsal striatum (i.e., putamen and caudate nucleus). Langen et al. (2011) claimed that the sensorimotor circuit is primarily involved in repetitive motor movements such as repetitive hand movements based on both animal and human studies. Lewis et al.'s (2007) animal study examined the role of cortical-basal ganglia circuity on repetitive behaviours by administrating pharmacological agents to cortical and subcortical areas within the sensorimotor circuit. In this study, deer mice were reared in two conditions (enriched and standard cages) and then classified as either "low stereotype" or "high stereotype," which thus formed four distinct groups: enriched low stereotype, enriched high stereotype, standard low stereotype, and standard high stereotype. The term "stereotypy" refers to repetitive motor movements such as hindlimb jumping and backward somersaulting. Pharmacological agents were then administered to block the cortico-striatal glutamatergic afferents and nigrostriatal dopamine projections. The enriched housed deer mice with low stereotypy exhibited noteworthy higher dendritic spine densities in the striatum and motor cortex compared with the other groups. Autism has been generally linked to dendritic spine dysgenesis which are believed to play a role in cognitive skills that are related to Executive Functioning (Martínez-Cerdeño, 2017; Phillips & Pozzo-Miller, 2015). The dendritic spine dysgenesis in autism that was found related to Executive Functioning skills and repetitive behaviours suggest a relationship between Executive Functioning and RRBs.

Structural magnetic resonance imaging (MRI) studies measuring the volume of brain structures in individuals with autism supported the previous assumptions and demonstrated that autistic individuals have increased volume of the putamen (Hollander et al., 2005; Langen et al., 2009) and reduced cortical thickness of primary sensory and cortical motor areas (Scheel et al., 2011; Shi et al., 2013). Hollander et al. (2005) examined the relationship of RRBs and the volume of caudate and putamen in a sample of 17 autistic participants (*M* = 28.4 years old) and 17 typically developing participants (*M* = 29.4 years old). Their caudate volume and putamen volume were measured with MRI and their RRBs were evaluated with the ADI-R. There was a significant relationship between caudate and putamen volume with higher-order RRBs (e.g., restricted interest and insistence on sameness) and but not with lower-order RRBs.

The above studies point to the relationship between the sensorimotor circuit and repetitive behaviours. In animal studies, this circuit was mainly correlated to repetitive motor movements, which is also known as “lower-order” RRBs as termed by Turner (1999). These findings were not surprising because the sensorimotor circuit has generally been related to sensory-motor repetitive movements. In human studies, the brain structures in this circuit were correlated to “higher-order” RRBs (e.g., restricted interest and insistence on sameness) but not to “lower-order” RRBs. However, it is important to note that the participants in Hollander et al.’s (2005) study were above 17 years of age (the exact age range was not shown in the article) and the 17 participants with ASC were verbal with IQ ranged from 60 to 127, whereas the 17 participants with typical development had IQ ranged from 88 to 122. Despite the wide range of IQ, the number of participants and their IQ range were not stated clearly, which can be crucial information because the types of repetitive behaviours and IQ have been found correlated (Willemsen-Swinkels et al., 1998).

Furthermore, the results collected from the ADI-R showed higher mean for higher order repetitive behaviours (M = 4.58) compared to lower order repetitive behaviours (M = 3.08). Therefore, it can be implausible to claim that sensorimotor circuit is not significantly correlated to “lower-order” RRBs if the study has limited number of participants exhibiting lower order behaviour.

### 1.9.2. “Cognitive” Circuit

The associative or “cognitive” circuit consists of the lateral orbitofrontal cortex (lOFC) and the dorsolateral prefrontal (dlPFC). The Orbitofrontal cortex (OFC) plays a vital role in Executive Functioning such as goal-directed behaviours (Fettes et al., 2017) and response inhibition (Bryden & Roesch, 2015) whereas the dlPFC is typically associated with working memory (Curtis & D’Esposito, 2003), response inhibition (B. L. Miller & Cummings, 2007), and cognitive flexibility (Kaplan et al., 2016). Executive Functioning and their role in ASC and RRBs are discussed in more detailed in section 1.3. Disintegrations (or disruption in the communication and coordination) in the cognitive circuit have been associated with rigidity, inflexible and ritualistic behaviours which were conceptualised as “insistence on sameness” repetitive behaviours (Geurts et al., 2009; South et al., 2007; Yerys, 2015). Similar to the sensorimotor circuit, these cortical regions (i.e., lOFC and dlPFC) project predominantly to the dorsal striatum (i.e., putamen and caudate nucleus). Lewis et al. (2007) examined the relationship between cognitive flexibility, an aspect of executive functions, and motor repetitive movements with an animal study. Deer mice were grouped into low and high repetitive behaviours and they were given a procedural learning task involving them turning to the left or right arm of a T-maze for reinforcement. Deer mice with high repetitive behaviours had more difficulties in reversing the direction from left to right, and vice versa for reinforcement in the T-maze, suggesting that cognitive inflexibility or rigidity were related to repetitive behaviours. This finding was perhaps predictable given that cognitive rigidity or deficits in cognitive flexibility share the common mediation by cortical-basal ganglia pathways with repetitive behaviours. Here again, we observe the potential relationship between cognitive flexibility and repetitive behaviours in the neurobiological account of RRBs.

### 1.9.3. "Emotion" Circuit

The limbic or "emotion" circuit is generally responsible for behavioural and emotional responses. The cortical regions in this circuit include the anterior cingulate cortex (ACC) and the orbitofrontal cortex (OFC). These regions are activated during the presence of rewarding or aversive stimuli from the environment (Kohls et al., 2014). In contrast to the sensorimotor and associative networks, the cortical regions project mainly to the ventral part of the striatum, which consists of the nucleus accumbens, olfactory tubule, ventral putamen, and ventral caudate. Then, the ventral striatum modulates the processing in the dorsal striatum, allowing cognitive and emotional information to direct sensorimotor behaviours. An imbalance between the ventral and dorsal striatum results in repetitive behaviours (Groenewegen et al., 2003). It has also been found that the volume of striatum components, particularly caudate, positively correlates to the degree of repetitive behaviours (Hollander et al., 2005) and negatively correlates with Executive Functioning (Voelbel et al., 2006). The relationship between performances on executive function tasks seems related to the brain structures of the limbic circuit which might explain RRBs (Voelbel, 2005). We will discuss whether there is any overlapping between the brain areas in this circuit and the brain areas being activated during emotional contexts in section 1.4.

The neurobiological account of RRBs, especially the cognitive and emotion circuits, prompted researchers to investigate the relationship of Executive Functioning and emotion aspect with RRBs. Although Yerys' (2015) review on the tripartite model suggested the consideration of alternative model such as cortico-striatal circuit as most of the MRI studies provided evidence supporting the role of cortico-striatal circuits in RRBs but not the thalamus and pallidum. The lack of evidence on the role of thalamus and pallidum is mainly due to the spatial resolution of MRI acquisition sequences for structural and functional MRI making it challenging to parse nuclei within these two regions. Regardless of the controversies regarding whether cortico-striatal circuits are related to RRBs through uni- or multi-dimensional effects, it is clear that the underlying neurobiological evidence relating to RRB presentation should be considered alongside evidence relating to Executive Functioning and emotion.

#### 1.9.4. Overlapped Brain Activations during ER tasks and RRBs

As part of the typical development trajectory, RRBs that appear during the first year of life reduce significantly by 4 years of age (Uljarević et al., 2017). The decline of these behaviours coincides with the manifestation of Executive Functioning skills (Leekam et al., 2011) which develop most rapidly between ages 3 to 5 years. Turner (1997) suggested a causal relationship between Executive Functioning and RRBs, but from the developmental trajectory account, Leekam et al. (2011) doubted the plausibility of a causal role of Executive Functioning on RRBs as these behaviours manifested at the very beginning of typical development. Leekam and colleagues stressed the importance of tracing the neurobiological changes alongside the occurrence of RRBs and the development of cortico-striatal circuits in typical early development as these immature movements are likely to be part of typical development. According to Thelen (1979), the high prevalence of repetitive motor movements in the first year of life is likely caused by slow maturation in cortical regions when the motor movements are not yet under voluntary control. They become increasingly under control towards the end of the first year when infants begin to develop more goal-directed behaviours, and consequently, their repetitive movements become more varied. Although the developmental account continues to stress the importance of cortico-striatal circuits in RRBs and argues that Executive Functioning deficits are not necessary in explaining RRBs, it fails to consider higher-order RRBs, such as restricted interest, insistence on sameness, and ritualised behaviours.

Higher-order RRBs reach their peak between 2 to 5 years old (Çevikaslan et al., 2014) and then decline from 8 to 14 years old in typical development (Zohar & Bruno, 1997; Zohar & Felz, 2001a). The developmental account struggles to explain the manifestation and reduction of higher-order RRBs and their functions. Iversen & Lewis' (2021) meta-analysis highlighted the importance of reopening the Executive Functioning account when explaining RRBs. They conducted three highly powered random-effects analyses on a sample size of 2964 to examine the strength of the relationship between RRBs and performance on set shifting (also known as cognitive flexibility), response inhibition, and Executive Functioning parental-report measures. These analyses reported a significant relationship between high RRB levels and 1) low performance in cognitive flexibility tasks ($r$ = 0.31, $p$ < .001), 2) low performance in response inhibition tasks ($r$ = 0.21, $p$ = .02) and 3) parental-report Executive Functioning measures ($r$ = 0.33, $p$ < .03). In response to Leekam et al.'s (2011) call to prioritize the cortico-striatal circuits when investigating RRBs, we hereby quote Langen et al. (2011) that "cognitive models have provided valuable hypotheses for how neurobiological circuitry might be disturbed in repetitive behaviour" (p.2). Therefore, we believe that both cognitive and neurobiological approaches should be considered, given that they are related.

In general, the developmental account overlooked the overlapping in the brain activation during Executive Functioning tasks and RRBs. Moll et al. (2002) examined the brain activation of seven typically developing men aged between 19 to 43 years during a trait making test (TMT), a neuropsychological test widely used to measure cognitive flexibility (Butler et al., 1991). It is worth mentioning that the researchers used a verbal adaptation of the TMT (vTMT) to make the most of the cognitive flexibility properties of the TMT while minimizing the visuomotor and visuospatial properties of the written TMT. The functional magnetic resonance imaging (fMRI) results revealed increased activation observed in the dorsolateral prefrontal cortex (dlPFC), supplementary motor area/cingulate sulcus, and intraparietal sulci during the vTMT task. As discussed previously in section 1.2.3, the dlPFC is an important region in the cognitive/associative circuit that has been associated to rigidity and ritualistic behaviours, and the cingulate sulcus also plays a critical role in the emotion/limbic circuit used to explain RRBs. Blasi et al. (2006) observed increased activation in the dlPFC, the ventrolateral prefrontal cortex (vlPFC), the dorsal cingulate (dACC), and the parietal cortex (PC) when 57 typically developing adults ($M$ = 28.6 years old) performed a modified version of the flanker task. The flanker task (Eriksen & Eriksen, 1974) was designed to measure the ability to suppress responses that are inappropriate in a given context (response inhibition). Other studies also found increased activation in dlPFC during long- and short-delay tasks (Barch et al., 1997), sentence reading and word retaining (Bunge et al., 2000), and increased activation in left inferior frontal cortex (IFC) and anterior cingulate cortex (ACC) during the reading span test (RST; Osaka et al., 2004). According to these studies, brain regions such as prefrontal cortex and cingulate cortex, have often been related with RRBs, activation in these regions during Executive Functioning tasks put forward the potential relationship between RRBs and Executive Functioning.

Thakkar et al. (2008) investigated the relationship between ACC activation during an anti-saccade task and repetitive behaviours in a sample size of 14 control ($M$ = 27 years old) and 12 participants with ASC (*M* = 30 years old). Participants with ASC displayed abnormally increased activation in ACC during correct trials alongside reduced fractional anisotropy in rostral ACC white matter. These activations were related to higher Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994) RRBs scores. Interestingly, Shafritz et al. (2008) found RRB symptoms *negatively* correlated with the activation in ACC and left intraparietal sulcus. The differences between Thakkar et al. (2008) and Shafritz et al.'s (2008) results could be due to task requirements, with participants in Thakkar et al.'s (2008) study required to inhibit responses whilst Shafritz et al. (2008) asked participants to do a cognitive flexibility task. Restricted and repetitive behaviours have also been found to be positively correlated with dlPFC activation. Ecker et al. (2013) investigated regional differences in cortical volumes, cortical thickness and surface area between 84 adults with ASC (*M* = 26.58 years old) and 84 control adults (*M* = 28.5 years old) with magnetic resonance imaging. Increased volume in dlPFC was also positively correlated with ADI-R communication domain measures and the ADI-R total score. ADI-R repetitive score, specifically circumstance interest domain, was also correlated with orbitofrontal cortex (OFC; Hardan et al., 2006) which plays a critical role in response inhibition tasks (Bryden & Roesch, 2015). From these studies, we know that the past studies have found brain regions that were involved in RRBs overlapped with the brain regions involved in the Executive Functioning tasks.

As discussed in the previous section, the basal ganglia, particularly the striatum, serves as a mediator for the pathways between the cortical areas to the motor cortex. These pathways are known as cortico-striatal circuits, which have been used to explain RRBs in ASC. The development rate of striatal volume (e.g., caudate, putamen and nucleus accumbens) were higher in ASC than typically developing children (Langen et al., 2009, 2014; Uddin, 2011). Langen et al. (2014) conducted a longitudinal structural magnetic resonance imaging scans with 49 children with ASC and 37 typically developing children, with a mean scan interval time of 2.4 years. The result found right caudate volume increased 4.6% in autistic children and 2.3% in typically developing children between the first scan (children aged 9.9 years on average) and the second (aged 12.3 years). This striatal growth for caudate, putamen and nucleus accumbens was positively correlated with ADI-R circumscribed interest domain; caudate and putamen volume correlated positively with ADI-R insistence on sameness. Caudate volume and putamen volume were also positively correlated with ADI-R circumscribed interest and insistence on sameness when combined (Hollander et al., 2005). Other studies have also found RRB symptoms positively correlated with the volume of putamen in individuals between 12 to 23 years old (Kenworthy et al., 2013), and caudate in 31 years old adults (Daly et al., 2014), children of age of 2 years (Wolff et al., 2014) and 12 years (Langen et al., 2009), and both adults and children between 7 to 44 years old (Rojas et al., 2006). All the studies focused on neuroanatomical structures and neurocircuits have found overlap between Executive Functioning and RRBs which highlights the importance of considering Executive Functioning when studying RRBs in ASC. However, there are a fair amount of studies that did not find significant correlations between RRB symptoms and the cortico-striatal circuits (Cascio et al., 2014; Kohls et al., 2012; Misaki et al., 2012; Shi et al., 2013; Yerys et al., 2015), as well as Executive Functioning (Boyd et al., 2009; C. R. G. Jones et al., 2018; Perry et al., 2022). These findings draw our attention to the possibilities of other factors affecting the relationship between Executive Functioning and RRBs.

### 1.9.5. Overlapped Brain Activations of ER, EF and RRBs

Reappraisal is one of the most commonly used adaptive Emotion Regulation strategies, altering behaviours and responses by reinterpreting the emotion-eliciting stimuli or situation (Gross, 2014). Based on some neuroimaging studies, successful reappraisal involves brain areas such as amygdala (Phelps & LeDoux, 2005), ventral striatum (Haber & Knutson, 2010) and insula (Uddin et al., 2014). These three brain regions are believed to be modulated via recruitment of a circuit of regions comprising the dlPFC, vlPFC, posterior medial prefrontal cortex (mPFC), posterior parietal cortex (PPC), and ACC (Martin & Ochsner, 2016). As described in the earlier section, the dlPFC is also commonly activated in tasks involving working memory (Bunge et al., 2000; Curtis & D'Esposito, 2003), cognitive flexibility (Kaplan et al., 2016; Moll et al., 2002b) and response inhibition (Blasi et al., 2006; B. L. Miller & Cummings, 2007). This region is also part of the associative circuit used to explain RRBs. vlPFC is commonly activated during tasks involving response inhibition (Blasi et al., 2006), an aspect of Executive Functioning that can be important in selecting a socially appropriate reappraisal strategy (Aron et al., 2004). ACC is generally active during tasks involving working memory (Osaka et al., 2004) and response monitoring (Shenhav et al., 2013), which can be important in being aware of own emotional states and regulate when necessary. Activation in ACC was also observed during saccade tasks related to RRBs (Shafritz et al., 2008; Thakkar et al., 2008). A summary of the overlapped brain structures across EF, ER and RRBs in ASC is presented in Table 1.1.

Table 1.1

*Summary Table of MRI Studies of Brain Region across Behaviours involving Executive Functions, Emotion Regulations and RRBs*

| Reference | Regions of interest | Participants | Methodology/Tasks | Findings |
|---|---|---|---|---|
| Ecker et al. (2013) | OFC<br>PCC | n = 84 ASD (age M = 26.58 years);<br>84 TD (age M = 28.5 years) | WASI (verbal, performance, full)<br>ADI-R (social, communication, RRBs)<br>ADOS<br>MRI scans | ASD group had higher cortical volume within frontal cortex regions and reduced surface area in OFC and PCC than control group. |
| Thakkar et al. (2008) | ACC | n = 12 ASD (age M = 30 years);<br>14 TD (age M = 27 years) | Antisaccades task<br>ADI-R (RRBs)<br>fMRI scans<br>Diffusion Tensor Imaging (DTI) | ASD group showed abnormally increased activation in rostral ACC (rACC) on correct trials.<br>Increased activation in rACC white matter on correct trials were related to higher levels of RRBs. |
| Shafritz et al. (2008) | ACC<br>dlPFC | n = 18 HFA (age M = 22.3 years); | ADI-R (RRBs)<br>Target detection task | ASD group showed reduced activation in dlPFC, IPS, basal ganglia (BG) regions |

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|---|---|---|---|---|
| | IPS<br>BG | 15 TD (age M = 24.3 years)<br><br>ASD group<br>WASI verbal IQ M = 103.3; performance M = 101.1, full M = 102.5;<br>TD group<br>WASI verbal IQ M = 111.2; performance M = 109.3, full M = 111.4 | fMRI scans | during target trials relative to control group.<br>ASD group showed reduced activation in dlPFC, ACC, IPS, BG regions during target-shift trials relative to control group.<br>For ASD group, activation in ACC and IPS regions negatively correlated with RRBs. |
| Hardan et al. (2006) | FC | n = 17 ASD (age = 8-12 years);<br>14 TD (age = 8-12 years) | MRI scans | For ASD group compared to TD, increased in total cerebral sulcal, gyral, temporal, and parietal thickness were observed but not in frontal and occipital cortex. |

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|---|---|---|---|---|
| | | | | No significant correlation between cortical thickness and ADI-R and ADOS scores for both groups. |
| Langen et al., 2009 | Striatum – caudate nucleus | n = 99 HFA (age = 6 – 25 years);<br>89 TD (matched) | Longitudinal study over 8 years<br>MRI scans<br>ADI-R items for repetitive motor behaviour, IS and circumscribed interests | Increased rate of growth higher in children with ASD not accounted for by overall changes in brain.<br>Caudate volume increased with age in ASD whereas it decreased with age in TD.<br>Caudate volume showed negative correlations with Insistence of Sameness, but not when controlled for age – correlation is more apparent in the younger half in a median split. |
| Hollander et al., 2005 | Striatum - Dorsal Striatum | n = 17 HFA (age = 17 - 55 years; IQ 60 - 127);<br>17 TD (age = 20.6 - 57 years; IQ 88 - 122) | MRI scans | Right caudate volume controlled for total brain volume larger for ASD group.<br>Right caudate and putamen volumes correlated with circumscribed interests and Insistence of sameness. |

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|---|---|---|---|---|
| Daly et al., 2014 | Inferior frontal cortex<br>Thalamus<br>Caudate<br>Cerebellum | n = 14 ASD (age *M* = 31 years);<br>14 TD (age *M* = 31 years) matched for gender, age and intelligence | Trial of acute tryptophan depletion;<br>fMRI scanning<br>Go/No-Go task | No depletion: For ASD group, reduced activation in inhibitory regions of Inferior frontal cortex and thalamus and increased activation of caudate and cerebellum.<br>Depletion of tryptophan: For ASD group, upregulated fronto-thalamic activation; downregulated striato-cerebellar activation.<br>Severity of autism related to differential modulation of activation by depletion. |
| Rojas et al. (2006) | Caudate nuclei, frontal and temporal regions, areas of cerebellum | n = 24 ASD (age = 7-47 years);<br>23 TD (matched age) | MRI scans | Partial associations between volumes of caudate nuclei, of multiple frontal and temporal regions, and of the cerebellum, and repetitive behaviours, after controlling for total grey matter volume. |
| Kenworthy et al., | Premotor regions of | n = 17 HFA (age = 12- | Verbal fluency | For ASD group, reduced neural response |

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|---|---|---|---|---|
| 2013 | FC<br>Thalamus<br>Putamen | 23 years);<br>20 TD (age = 12-21 years) | (automatic speech, category fluency and letter fluency conditions)<br>fMRI scans | than TD in multiple regions of left anterior and posterior cortices, and sub-cortical structures, including three premotor regions in frontal cortex, bilateral thalamus and putamen.<br>Activity in putamen and thalamus negatively correlated with autism repetitive behaviours in ASD group. |
| Cascio et al., 2014 | ACC<br>Insula | n = 19 ASD (age *M* = 12.58);<br>18 TD with strong interests/hobbies (age *M* = 13.11 years) | Parent-report of interest<br>Operant task<br>fMRI scans | Increased BOLD response in affective neural regions for both groups.<br>Increased BOLD response in insula and ACC in ASD compared to TD.<br>Parent report of intensity and interference with daily life predicted insula response. |
| Misaki et al., 2012 | OFC<br>dFC | n = 41 ASD (age 12-24 years; *M* = 16.75 years); | Kernel Canonical Correlation analysis on Whole brain cortical | Thinner cortex for ASD.<br>For TD group, IQ correlated to cortical thickness in OFC, postcentral and |

| | | | | |
|---|---|---|---|---|
| | | 40 TD (age 12-23 years; *M* = 17.04 years) | thickness with MRI | superior temporal regions and greater thinning with age in dorsal frontal cortex (dFC) in IQ >120;<br>these associations were not seen in ASD. |
| Shi et al., 2013 | CC<br>FC | n = 49 ASD (age = 6-15 years, *M* = 9.6 years);<br>51 TD (age = 6-15 years, *M* = 9.7 years) | MRI scans | For ASD group, increased intra- and inter-module connectivity in middle frontal gyrus, inferior parietal gyrus, and cingulate cortex (CC); increased correlation strengths between regions inside frontal cortex, impaired correlation strength between frontotemporal and frontoparietal regions, relative to TD. |
| Yerys et al., 2015 | PFC<br>Striatum<br>PC<br>Cerebellum | n = 20 ASD (age = 7-14 years);<br>19 TD (matched age) | Set shifting task<br>MRI scans | Activation in PFC, striatum, PC, and cerebellum in both groups during task.<br>For ASD group, decreased activation of right middle temporal gyrus across all trials, increased activation of mid-dorsal cingulate cortex/superior frontal gyrus, |

| | | |
|---|---|---|
| | | left middle frontal, and right inferior frontal gyri during the Switch vs. Stay contrast. |

## 1.10. EF, ER and RRBs

Both cognitive and neurobiological models provide valuable information while investigating the relationship between Executive Functioning and Emotion Regulation as well as their potential roles in RRBs.

Firstly, many studies found a positive relationship between Executive Functioning and Emotion Regulation in typically developing children (Carlson & Wang, 2007; Li-Grining, 2007; McRae, Gross, et al., 2012; Schmeichel et al., 2008b). A causal relationship was suggested when Li et al.'s (2020) study reported executive function training improves children's ability in regulating their emotion. However, it is important to note that studies that examined the relationship between executive function and emotion regulation were only limited to typically developing samples.

Executive Functioning and Emotion Regulation have both been related to RRBs in both typical development and ASC. In typical development, as Executive Functions (e.g., response inhibition) manifest and improve across 2 to 4 years old (Posner & Rothbart, 1998), a decline of RRBs can be observed (Leekam et al., 2011). Executive dysfunctions have been used to explain the challenges in ASC and the occurrences of RRBs. For example, Iversen and Lewis' (2021) meta-analysis found significant negative relationships between RRBs and Executive Functions, such as cognitive flexibility and response inhibition. Autistic children with weak Executive Functioning usually have more RRBs.

In addition to Executive Functioning, studies that examined the potential role of RRBs as an emotional regulatory mechanism have consistently found autistic children with anxiety displayed more RRBs than autistic children without anxiety (Joyce et al., 2017; Lidstone et al., 2014; Muskett et al., 2019; Rodgers et al., 2012; K. M. Russell et al., 2019).

RRBs were observed during other emotional contexts too (Hirschler-Guttenberg et al., 2015; Willemsen-Swinkels et al., 1998).

We understand from the past studies that Executive Functioning relates to RRBs, and Emotion Regulation also relates to RRBs in autistic population. A positive correlation and a causal relationship were observed between Executive Functioning and Emotion Regulation in typically developing individuals, but no studies have investigated this relationship in autistic population. Through mediation analysis, can Emotion Regulation be identified as a significant mediator in the relationship between Executive Functioning and RRBs in individuals with ASC? In light of prior research findings, we hypothesize that Emotion Regulation serves as a significant mediator in the relationship between Executive Functioning and RRBs in individuals with ASC, indicating that the influence of Executive Functioning on RRBs is partially or fully mediated by the regulation of emotional processes.

Additionally, we are also interested to learn how do different emotional contexts influence the occurrence of RRBs in individuals with ASC, and to what extent do these contexts interact with Executive Functioning and Emotion Regulation? We hypothesize different emotional contexts significantly impact the occurrence of RRBs in individuals with ASC, and there is a significant interaction between emotional contexts, Executive Functioning, and Emotion Regulation, suggesting that RRBs manifest differently depending on the emotional environment.

As mentioned previously, children with ASC and their families often experience challenges in their everyday life resulted from RRBs, executive dysfunction and emotion dysregulation. We believe that further understanding the relationship between these constructs is necessary to further our theoretical understanding of the challenges related to them. At the time of writing this thesis, no published studies have looked at this two-way relationship. Therefore, in this thesis we will examine the relationships between these behaviours in children with ASC and typically developing children.

# 2 Chapter Two: Emotion Regulation: Mediator between Executive Functioning and Restricted and Repetitive Behaviours (RRBs)

## 2.1 Introduction

Executive dysfunction and emotion dysregulation have been consistently observed in individuals with Autism Spectrum Conditions (ASC), and existing studies shed light on the potential roles of these two constructs with regard to Restricted and Repetitive Behaviours (RRBs). In this chapter, I will further explore how Executive Functioning and Emotion Regulation are related to RRBs in ASC with a systematic review and an experimental study.

### 2.1.1 Systematic Literature Search

This chapter aims to examine the relationship between Executive Functioning and Emotion Regulation, and their effects on RRBs across children with ASC and typical development (TD). In this introduction, a systematic search of published literature was conducted to identify and evaluate the existing literature on Executive Functioning and Emotion Regulation and their relationship with RRBs. The articles that meet the searching criteria will be discussed. All published literature available to July 2021 was identified according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al., 2009). The electronic databases MEDLINE, Web of Science, PsycINFO, Cochrane, and Education Resources Information Center (ERIC) were searched concurrently for entries containing any combination of the following terms in the Title, Abstract, or Keyword search fields:

1. “Autism” or “Autis*” or “Asperger*” or “ASD” or “Pervasive Developmental Disorder” and

2. “repetitive behav*” or “repetitive movement*” or “stereotypic movement*” or “stereotyp*” and
3. “executive function*” or “set shifting” or “working memory” or “inhibition” or “inhibitory control” or “cognitive flexibilit*” and
4. “emotion* dysregulation” or “emotion* regulation” or “emotion* control” or “emotion* management”.

The initial search identified 1813 articles. Key journals such as Autism, Autism Research, Journal of Child Psychology and Psychiatry, Journal of Autism and Developmental Disorders, and Research in Autism Spectrum Disorders were hand searched. This search strategy identified another 116 articles. Of these 1929 papers, 34 duplicates were removed. The screening was conducted in three steps: (1) title screening, (2) abstract screening, and (3) full-text screening. The inclusion and exclusion criteria of the title screening were determined after searches were conducted. Articles were selected if the title included any terms related to (1) restricted and repetitive behaviours (stereotypical movements, restricted and repetitive behaviours (RRBs); (2) Executive Functions (cognitive, Executive Functioning, cognitive flexibility, set-shifting, inhibition, inhibitory control, working memory); and (3) emotion or Emotion Regulation (anxiety, excitement, anger, Emotion Regulation, emotion experiences, human emotion, emotion dysregulation). Articles were excluded if the title involved any terms related to (1) intervention, (2) animal experiments, (3) neurology and pharmacology, and (4) populations that do not include ASC.

The inclusion and exclusion criteria of the abstract screening were determined before searches were conducted. Articles were included if they met all of these criteria: (1) the target population had a diagnosis of ASC (i.e., Autism Spectrum Disorder, Autistic Disorder or Pervasive Developmental Disorder-Not Otherwise Specified) and might include other samples as one of the groups; (2) the participants had RRBs; (3) the participants' RRBs were measured; (4) the study had either Executive Functions or Emotion Regulation as one of the main variables; (5) the article was published in the English language and (6) was peer-reviewed. Articles were excluded if: (1) they were not available online; (2) they were not available in the English language; (3) they were not data-based (e.g., reviews, books, theoretical papers); or (4) the target population did not have a diagnosis of ASC. A summary of the selection procedure is presented in Figure 2.1. The selection of literature was conducted by the author and a Bachelor's student majoring in Psychology. The systematic search of the literature identified eight studies investigating the role of Executive Functions or Emotion Regulation in RRBs in ASC. A detailed description of the studies is provided in Table 2.1.

**Figure 2.1.**

*Literature selection diagram.*

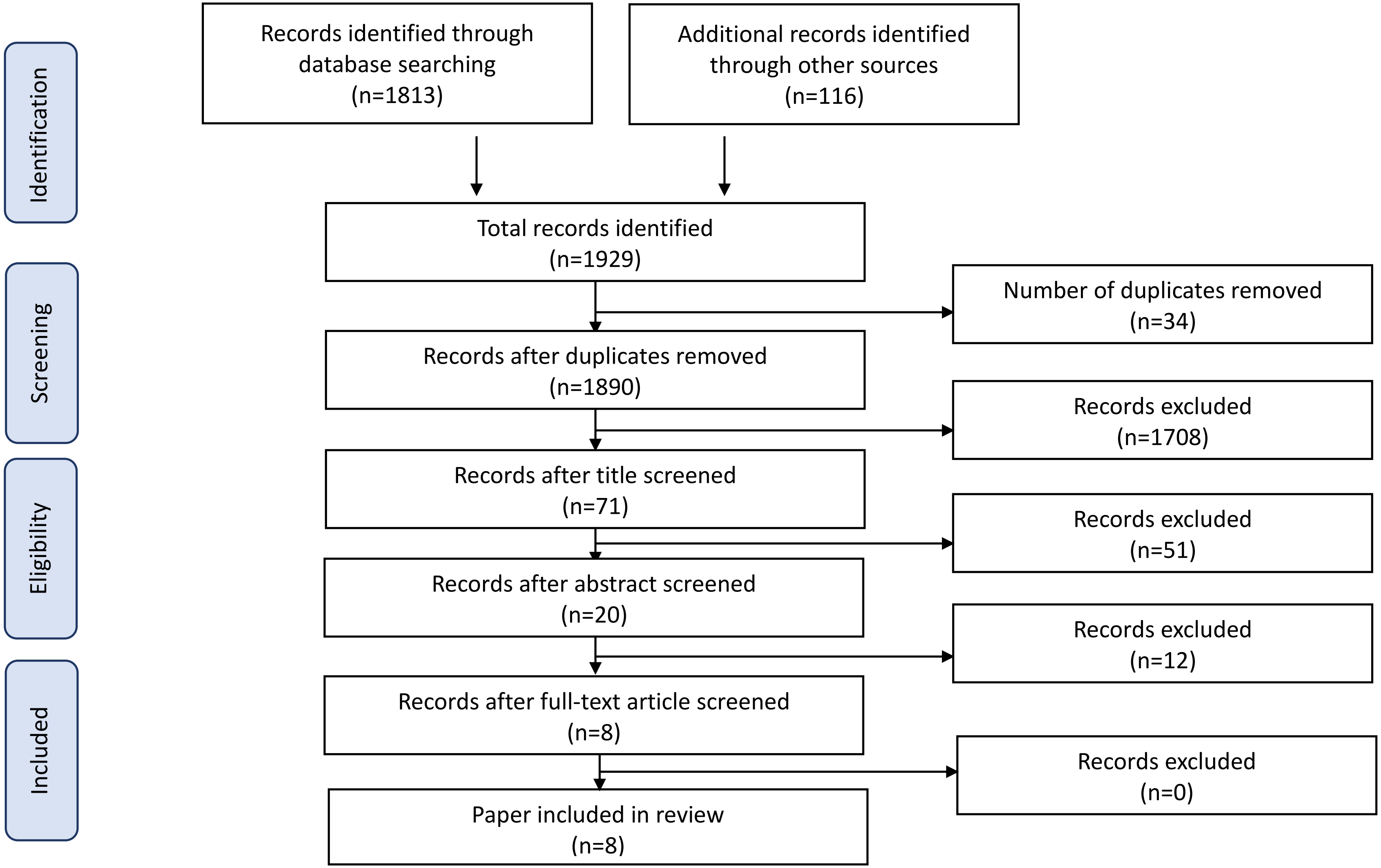

**Table 2.1.**

*Studies on the role of Executive Function (EF) or Emotion Regulation (ER) on RRBs in ASC.*

| Author (year) | Participants | EF Measure | ER Measure | RRBs Measure | Findings |
|---|---|---|---|---|---|
| Boyd et al. (2009) | *n* = 61 ASC (31 ASC, 22 AS, 5 PDD-NOS, 3 unspecified), 64 TD (age = 6-17 years); IQ ≥ 70; absence of medical issues and uncorrected visual impairment; ASC diagnosis confirmed by ADI-R. | BRIEF | Not Available (N/A) | RBS-R | BRIEF score was correlated with RBS-R subscales of self-injury, compulsions, rituals/sameness, and total score. Group, chronological age, sensory sensitivity (measured by SQ) and BRIEF behavioural regulation predicted RBS-R total score. |
| Kim & Lord (2010) | *n* = 121 ASC, 71 PDD-NOS, 90 NS, 173 TD (age = 8-56 months). NVIQ = ASC (*M* = 62.94), PDD-NOS (*M* = 72.5), NS (*M* = 77.18), TD (*M* = 113.11); Diagnosis given based | MSEL BSID | N/A | ADOS | Lower NVIQ (measured by MSEL and BSID) predicted higher RRBs for all groups across all ages. When analysed separately, NVIQ did not predict RRBs for ASC at any age, but NVIQ significantly predicted RRBs for |

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| | on DSM-IV | | | | PDD-NOS, NS, and TD above or equal to 25 months, but not for those below 25 months. |
| Cervantes et al. (2014) | *n* = 325 ASC, 1894 atypical (age = 17-39 months); Developmental Quotient (DQ) = 194 ASC with typical cognitive ability, 131 ASC with low cognitive ability, 1694 atypical with typical cognitive ability, 200 atypical with low cognitive ability | BDI-2 | N/A | BISCUIT-Part3 | Children with ASC (typical and low DQ) displayed significantly more repetitive behaviours than children with atypical development (typical and low DQ). Children with ASC (typical DQ) displayed more repetitive behaviours than children with ASC (low DQ). |
| Vasa et al. (2018) | *n* = 57 ASC (without co-occurring ID) and 32 TD, (age = 7 - 16 years); ASC diagnosis supported by ADI-R & ADOS; | N/A | BRIEF (Emotion Control) SCARED- | IUS-C RBS-R | Intolerance of Uncertainty (IU) was significantly correlated with BRIEF Emotion Control (ED) and repetitive behaviours but not with social |

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| | WISC-IV IQ ≥ 80 if not, VCI/PRI ≥ 80 or lower of two ≥ 65. | | C & SCARED-P | | communication deficits. ED predicted IU, but repetitive behaviours were not a significant predictor of IU, when controlling for anxiety, and all variables were entered together. |
| Bos et al. (2019) | *n* = 62 ASC and 38 TD, (age = 6 - 12 years); ASC diagnosis supported by ADOS-2; TD were screened for ASC with SCQ-Lifetime | Go/No-Go | SCQ | ADOS-2 RBS-R CBCL SRS | In the ASC group, the RBS-R score on the Ritualistic/Sameness factor negatively correlated with cognitive control to affective cues of interest. The correlation between cognitive control, affective cues, and other RBS-R factors were not significant. |
| Faja & Nelson Darling (2019) | 102 ASC (age = 7 – 11 years); ASC diagnosis confirmed using ADOS & ADI-R; WASI-2 IQ ≥ 80 & verbally fluent | Stroop task Change task BRIEF | N/A | ADOS-2 ADI-R | Stroop congruent-incongruent difference scores were related to higher-order RRBs but not lower-order (sensorimotor) RRBs. Stroop scores significantly predicted |

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| | | | | | higher-order RRBs when controlling for age and IQ.<br>Change Task stop signal reaction time (SSRT) was unrelated to higher-order or lower-order RRBs. Increased inhibition during Change Task was unrelated to higher-order RRBs but related to lower-order RRBs.<br>BRIEF Inhibit score and Shift score were positively related to higher-order RRBs but not lower-order RRBs. BRIEF Shift significantly predicted higher order when controlling age and IQ. |
| Ibrahim et al. (2019) | *n* = 63 ASC, 79 individuals with disruptive behavior disorder (DB), and 40 TD. (age | N/A | ARS | ADOS-2<br>SRS-2 | In the total sample, anger rumination was positively correlated with RRBs and aggressions. In the ASC group, anger |

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| | = 8-16 years); ASC diagnosis confirmed using ADOS & ADI-R; CBCL of a T score ≥ 65. | | | | rumination was positively correlated with RRBs but not for children without ASC. In the ASC group, ASC diagnosis predicted anger rumination. RRBs, but not aggression, predicted anger rumination. |
| Fernandez-Prieto et al. (2020) | *n* = 79 ASC (age = 4-16 years); diagnosis confirmed by ADI-R & ADOS. | N/A | CSP-2 | CBCL | Emotion regulation and control were positively correlated with almost all behaviours, including repetitive/obsessive behaviour. Working memory was positively correlated with repetitive/obsessive behaviour only. Emotion regulation and control were positively correlated with touch, movement, and body position sensory processing. Working memory was positively correlated with touch and movement. |

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| | Repetitive/obsessive behaviour was significantly correlated with sensory processing, including auditory, visual, touch, movement, and body position.<br>A mediation effect of Executive Functioning, specifically emotion regulation and control, was observed in the relationship between sensory processing and behavioural problems. |

Abbreviations: ARS, Anger Rumination Scale, BDI-2, Battelle Developmental Inventory, Second Edition, BISCUIT-Part3, Baby and Infant Screen for Children with Autism Traits – Part3, BRIEF, Behavior Rating Inventory of Executive Function, BSID, Bayley Scales of Infant Development, CBCL, Child Behavior Checklist, CSP-2, Child Sensory Profile 2, DAS-ll, Differential Abilities Scale-Second Edition, DQ, Developmental Quotient, IUS-C/P, Intolerance of Uncertainty Scale: Child and Parent Versions, MSEL, Mullen Scales of Early Learning, NS, NonSpectrum, PDD-NOS, Pervasive Developmental Disorder, Not Otherwise Specified, NVIQ, Nonverbal IQ, RPQ, Reactive-Proactive Aggression Questionnaire, SCARED-C/P, Screen for Childhood Anxiety Related Disorders: Child and Parent Versions, SCQ, Social Communication Questionnaire, SQ, Sensory Questionnaire, SRS-2, Social Responsiveness Scale, Second Edition, TD, Typically Developing, WASI-2, Wechsler Abbreviated Scale of Intelligence®, Second Edition.

Note NonSpectrum (NS) disorder consists of children with a history of developmental delay without a diagnosis of ASC.

Out of the eight studies on RRBs, only one focused on both Executive Functioning and Emotion Regulation. Three studies focused on emotion-related variables (e.g., Emotion Regulation, emotion control, and anger rumination), and four focused on cognition-related variables, such as intelligence quotient, development quotient and cognitive control, and behaviour regulation. These studies used instruments such as the Autism Diagnostic Interview-Revised (ADI-R), the Autism Diagnostic Observation Schedule (ADOS) or its Second Edition (ADOS-2), Child Behavior Checklist (CBCL), Intolerance of Uncertainty Scale: Child and Parent Versions (IUS-C/P), RBS-R, and Social Responsiveness Scale, Second Edition (SRS-2) to measure different aspects of RRBs. Instruments such as the Behavior Rating Inventory of Executive Function (BRIEF), two domains of the CBCL (i.e., working memory and planning), the Change task, Go/No-Go, and Stroop task, were used to measure Executive functions. Instruments that were used to measure emotional variables include the Anger Rumination Scale (ARS), Screen for Childhood Anxiety Related Disorders: Child and Parent Versions (SCARED-C and SCARED-P), and one domain of the CBCL (i.e., emotion regulation and control).

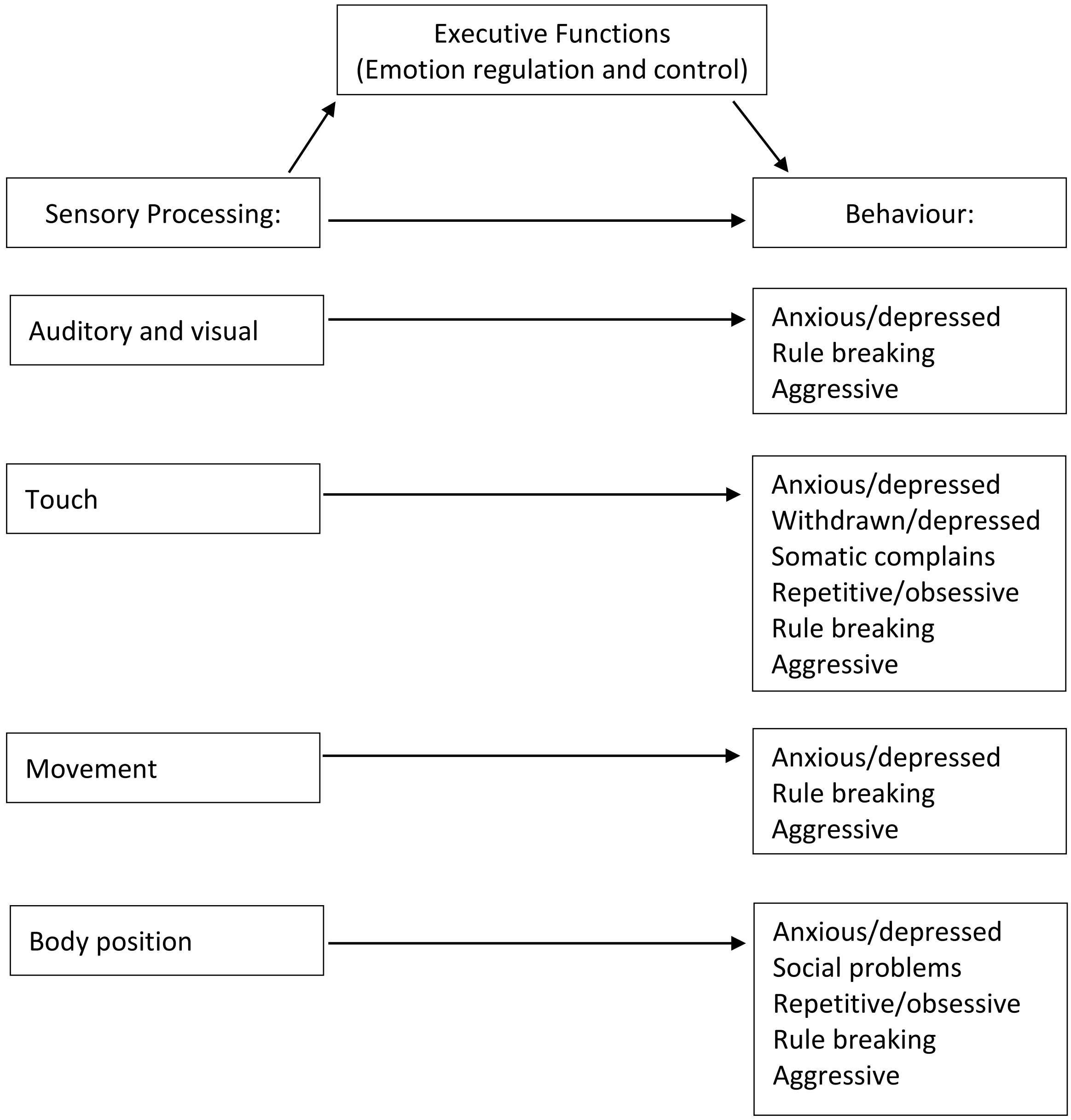
According to the systematic search, the study by Fernandez-Prieto et al. (2020) was the only study on RRBs that concurrently examined emotion regulation and control, together with constructs of Executive Functioning such as working memory and planning. It is crucial to note that Fernandez-Prieto and colleagues (2020) considered emotion regulation and control as a domain that belongs to Executive Functioning. Generally, Emotion Regulation and Executive Functioning have been discussed as two separate and independent domains in papers involving individuals with ASC (Carlson & Wang, 2007; Li et al., 2020; Li-Grining, 2007) and typically developing individuals (Garcia-Andres et al., 2010; Lantrip et al., 2016; Sudikoff et al., 2015). The main objective of the study by Fernandez-

Prieto et al. (2020) was to investigate the mediating effect of Executive Functioning in the relationship between four types of sensory processing and eight behaviours. Seventy-nine children and adolescents with ASC between the ages of 4 and 16 years (age *M* = 9.01 years, *SD* = 2.9 years) were recruited for this study. They received a diagnosis of ASC from a psychiatrist and a psychologist based on the DSM-5 criteria. CBCL was used to measure eight behaviours, including repetitive/obsessive behaviour and three Executive Functioning domains (emotion regulation and control, working memory and planning). The participants' sensory processing features were also examined with Child Sensory Profile-2, CSP-2. The results found that only repetitive/obsessive behaviour was associated significantly with working memory and almost all behaviours (i.e., anxious/depressed, rule breaking, aggressive, somatic complains, repetitive/obsessive, and social problems), except withdrawn/depressed, were significantly associated with the emotion regulation and control domain. Fernandez-Prieto et al. (2020) suggested that children with difficulties in temporarily retaining and manipulating necessary information (working memory ability) to execute cognitive tasks are more likely to be frustrated and engage with repetitive behaviours and those with difficulties in managing their emotions typically engaged in more behavioural problems (e.g., anxiety, isolation, aggressive or rule-breaking behaviours). Consistent with other studies (Li et al., 2020; Miyake et al., 2000), the findings from this study supported that emotional competencies are affected by Executive Functioning abilities.

Fernandez-Prieto et al. (2020) further analysed the data with structural equation modelling (SEM) methods. The mediating role that Executive Functions' emotion regulation and control domain play in the relationship between four sensory processing and specific behaviours are presented in Figure 2.2.

**Figure 2.2.**

*Fernandez-Prieto et al.'s (2020) diagram illustrating the mediating effect of Executive Functions (emotion regulation and control domain) in the relationship between sensory processing and behaviour.*




Atypical sensory processing has long been observed in individuals with ASC and is considered a defining characteristic of autism in the DSM-5 (American Psychiatric Association, 2013). As mentioned in Chapter 1, RRBs have been conceptualized as a sensory regulatory mechanism for individuals with ASC, in which they engage in sensory motor repetitive behaviours to seek for sensory inputs or use rigidity and ritualistic behaviours to avoid sensory stimulation. The CPS-2 was used to analyse sensory processing features in Fernandez-Prieto et al.'s (2020) study. However, although Fernandez-Prieto and colleagues (2020) aimed to examine the sensory processing as an independent entity, they used only one of the subscales (Sensory System subscale) to measure each sensory modality and its relationship with behaviours, rather than the total scale. In comparison with analysing scores from each sensory modality (i.e., auditory and visual, touch, movement, and body position) and its relationship with each behaviour, it is believed that a composite score will provide greatest increase in power (Song et al., 2013). Fernandez-Prieto and colleagues (2020). Secondly, the reliability of each sensory modality from this subscale was not examined in this study but previous studies had reported acceptable but low internal consistency alpha values ranging from .60 to .85 for the Sensory System subscale relative to the Sensory Pattern subscale from the CPS-2 ranging from .82 to .86 for the ASC population (Mirzakhani et al., 2021). Therefore, the usage of only the Sensory System subscale, given its low consistency and the reliance on one measure, it is possible that this construct is not adequately measured.

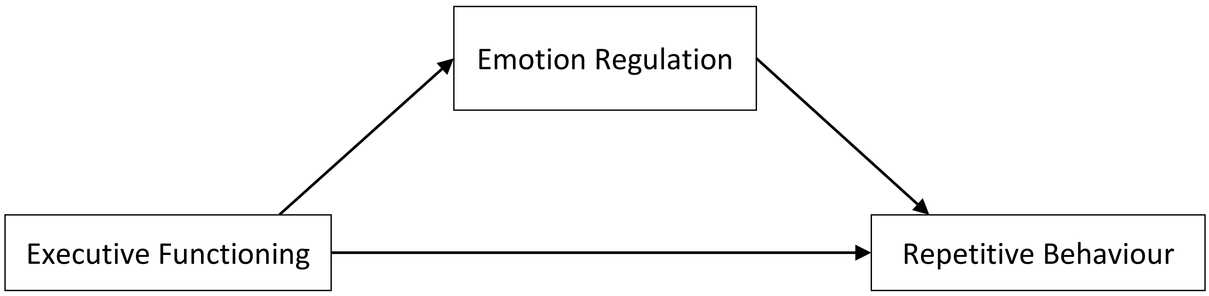
Fernandez-Prieto et al. (2020) also relied on CBCL to measure two main variables (Executive Functioning and behaviour) in the study. Warnick et al.'s (2008) systematic review reported that the CBCL had a moderate sensitivity of 66% (95% CI 60% to 73%) and specificity of 83% (95% CI 81% to 85%) for total problems (includes Internalizing, Externalizing, Thought, Social, and Attention Problems). No studies have investigated the sensitivity and specificity of the three Executive Functioning subscales in the CBCL questionnaire. Fernandez-Prieto et al. (2020) assessed Executive Functioning domains (emotion regulation and control, working memory, and planning) as suggested by McCray et al. (2014). Although moderate to strong reliability was reported for working memory (Cronbach's alpha = .74), and emotion regulation and control (Cronbach's alpha = .87), low reliability (Cronbach's alpha = .39) was reported for the planning domain and was thus removed from the analysis. Fernandez-Prieto et al. (2020) included only the emotion regulation and control domain as the Executive Functioning in their article and excluded other domains such as working memory and planning under the Executive Functioning domain. The authors further explained the findings that Executive Functions (through emotion regulation and control) affect how one processes sensory information and how one responds to the sensory stimulation with their behaviours. ASC children with atypical sensory processing are more likely to have emotional dysregulation which has a cascading effect on their behaviour. However, the authors also cited Miyake et al. (2000) acknowledging that emotional competencies "are influenced by Executive Functioning." It was unclear whether the authors were considering Executive Functioning and Emotion Regulation as a united or diverse entity.

Another plausible explanation of their findings is that Executive Functioning affects how individuals process sensory information, as proposed by Fernandez-Prieto et al. (2020) but instead of considering Executive Functioning (through emotion regulation and control) as an independent entity, individuals with poor Executive Functioning are more prone to use maladaptive emotion regulation strategies (Predescu et al., 2020) and these strategies can be mirrored in their behaviour. The significant relationship between Executive Functioning and Emotion Regulation has long been reported by many studies (Fuster et al., 2009; Gerardi‐Caulton, 2000; Li et al., 2020; Oswald, 2013; Predescu et al., 2020) and has also been discussed comprehensively in Chapter 1. Fernandez-Prieto et al. (2020) considered

Emotion Regulation as part of Executive Functioning, while based on the literatures, they could be considered two separate constructs, and hence in this thesis, we proposed a new relationship as presented in the following illustration (see Figure 2.3).

**Figure 2.3.**

*An illustration demonstrating the alternative relationship between Executive Functioning, Emotion Regulation and Repetitive Behaviour.*




### 2.1.2 Autistic Features and Restricted and Repetitive Behaviours (RRBs)

Apart from atypical sensory processing, scientists have also linked cognitive or motor abnormalities that co-occur with RRBs as a potential mechanism of RRBs. In the cognitive domain, the development of RRBs has been theorised as consequent to impairments in Executive Functioning (Evans et al., 2004; Lopez et al., 2005; Turner, 1997), theory of mind (Jones et al., 2018) and weak central coherence (Chen et al., 2009; South et al., 2007). As described in Chapter 1, deficits in Executive Functioning have been widely demonstrated in individuals with ASC particularly impaired cognitive flexibility (Ozonoff et al., 1991; Ozonoff & Strayer, 1997) and impaired inhibition of a prepotent response (Hill, 2004; Ozonoff et al., 1994). According to Lopez et al. (2005), deficiency in tasks that index cognitive flexibility was positively correlated to the degree of RRBs in ASC. Theory of mind (ToM), the ability to deduce and infer others' mental states and to use this information to predict their behaviours, has also been found to associate with RRBs. Jones et al. (2018) assessed the

ToM of 100 adolescents with ASC (*M* = 15 years 6 months, *SD* = 6 months) using four ToM tasks: False belief, Stranger stories, Frith-Happe animations, and Reading the mind in the eyes tasks (RMET) and their RRBs with the Repetitive Behaviour Scale-Revised (RBS-R). The results showed that all tasks indexing ToM were positively related to RRBs. Additionally, individuals with ASC show a more detail-focused cognitive processing style with greater emphasis and attention to local information, and struggle in tasks indexing central coherence which requires them to integrate details to derive global information (Happé & Frith, 2006). A significant relationship between the speed of completing the Embedded Figures Test (EFT), which is generally used to identify individuals' cognitive style, and the degree of RRBs (as measured by the Childhood Routines Inventory; CRI) was reported in high-functioning children with ASC suggesting that children who demonstrated a detail-focussed cognitive style tend to have more RRBs (Chen et al., 2009).

In the motor domain, there are reports of motor impairments in individuals with ASC including impaired imitation (Jones & Prior, 1985; Rogers et al., 1996), postural instability (Kohen-Raz et al., 1992; Molloy et al., 2003), and motor incoordination (Hallett et al., 1993). As described in Chapter 1, RRBs can be observed throughout the early stage of typical development and these behaviours are theorized to play an essential role in neuromuscular and motor development (Thelen, 1979). Children with ASC demonstrate delayed or atypical attainment of motor development milestones, such as showing asymmetry in arms and/or legs when crawling, or skipping the crawling stages (Teitelbaum et al., 1998). Markedly different postural movement profiles and impaired motor control were reported in children with developmental disabilities who display RRBs (specifically repetitive motor movements or stereotyped behaviours) relative to children without RRBs (Bodfish et al., 2001). This study further supported the relationship between motor impairments and RRBs.

In summary, autistic features such as executive dysfunction, weak ToM, detail-focused cognitive styles, and impaired motor functioning, in addition to atypical sensory processing, have been found significantly related to RRBs.

### 2.1.3 Objectives

There are three objectives in this chapter. Firstly, Executive Functioning, Emotion Regulation and RRBs will be examined across children with ASC and TD children. Secondly, the relationship between Executive Functioning and Emotion Regulation will be examined. Previous research has observed a significant positive correlation (Schmeichel et al., 2008a) and causal relationship (Ferrier et al., 2014; Li et al., 2020) between Executive Functioning and Emotion Regulation in TD children. Although no research to date has specifically investigated these relationships in children with ASC, it is expected that children with better Executive Functioning can regulate their emotions better. Finally, the role of Emotion Regulation as mediator in the relationship between Executive Functioning and RRBs will be investigated using Hayes' PROCESS model 4. Emotion Regulation is postulated to mediate the relationship between Executive Functioning and RRBs (see Figure 2.3 in Results for reference).

As described in Chapter 1, significant relationships between Executive Functioning and Emotion Regulation, Executive Functioning and RRBs, as well as Emotion Regulation and RRBs have been observed independently but no studies have looked at the two-way relationship between them. Based on past research, we expect to observe Emotion Regulation mediating the relationship between Executive Functioning and RRBs. This chapter aims to examine this relationship in children with ASC and typical development.

## 2.2 Method

### 2.2.1 Challenges during the Covid-19 Pandemic

The initial intention of this Chapter was to assess Executive Functioning domain with neuropsychological tasks and RRBs with on-site observation. However, due to the Covid-19 pandemic and concomitants travel and social restrictions, the study was amended such that it could be delivered via an online platform.

### 2.2.2 Participants

The sample for this study comprised 64 participants which 31 parents with a typically developing child, aged 4 years to 13 years (*M* = 8 years 3 months, *SD* = 3 years), and 33 parents with a child with ASC, aged 4 years to 13 years (*M* = 7 years 10 months, *SD* = 2 years 9 months). The sample is selected based on the schooling ages in Malaysia. Preschool education in Malaysia typically starts for children at the age of 4 or 5, depending on the family's or parents' preferences. Primary school education concludes by the age of 12. In this study, we recruited children from 4 to 13. We included children of the age of 13 considering some children from the public primary schools with an integrated special education programme, namely Program Pendidikan Khas Integrasi (PPKI), in Bahasa Malaysia, entered the primary school integrated programme one year later than their peers.

Six parents from the latter group were excluded from the data analysis as their child has other disorders besides ASC, such as attention deficit hyperactivity disorder (ADHD), sensory processing disorder (SPD), and social communication delay. The remaining 27 parents' children had a clinical diagnosis of autism spectrum conditions.

Diagnosis: Based on the parent reports, each child from the ASC group had a clinical diagnosis by a professional such as medical practitioner, paediatrician, family medicine specialist, or psychiatrist. These professionals are recommended diagnosis providers by the

Ministry of Health Malaysia in the Clinical Practice Guidelines for Management of Autism Spectrum Disorder in Children and Adolescents (Malaysian Health Technology Assessment Section, 2014). We did not use international assessments, such as ADOS-2 and ADI-R, to reassess the children's diagnosis as, to the best of our knowledge, these assessments have not been appropriately validated for this sample. We used the Autism Spectrum Quotient (AQ-10; Allison et al., 2012) as a measure of autism traits to quantify differences between TD and ASC groups.

Parents were recruited via two methods: 1) an invitation brochure was shared on social media platforms, and 2) an invitation email was sent out to mainstream tuition centres, and public and private special education centres which provide intervention and assessment services to children with ASC. They were screened for eligibility during the first contact. In the second contact, the information sheet and consent form were presented and explained to the parents. The questionnaire link was then shared with those who met the inclusion criteria. Inclusion criteria for the TD group were the following: the child, aged 5 to 13 years old, had no formal diagnosis nor suspected of any developmental, neurological, or psychiatric disorders or a known genetic condition, such as Down Syndrome. Inclusion criteria for the ASC group were that the child was aged 5 to 13 years old, had received a formal diagnosis of ASC by a professional (e.g., medical practitioners, paediatricians, family medicine specialists, or psychiatrists), with no brain injury, epilepsy, or known genetic condition.

#### 2.2.2.1 Ethics

This study has been reviewed by the University of Reading Malaysia Research Ethics Committee and has been given a favourable opinion for conduct with *UoRM REC 2020/01* as the unique approval reference number. All researchers working on this project have had the appropriate criminal records checked by the Ministry of Foreign Affairs and received a Certificate of Good Conduct.

### 2.2.3 Materials and Measures

#### 2.2.3.1 Autistic Traits Measure

The *Autism-Spectrum Quotient (AQ-10;* Allison et al., 2012) is a ten-item brief screen for ASC based on the AQ-50 (Baron-Cohen et al., 2001). Each item is answered on a four-point scale, ranging from “definitely agree” to “definitely disagree”. The “disagree” responses are scored 0, and the “agree” responses are scored 1 for half of the items and vice-versa for the other half. The total score is from 0 to 10. The internal consistency of the total score for AQ-10 (Child Version) was .90 (Allison et al., 2012). The AQ-10 (Child Version) significantly correlated with the AQ-50 (Child Version) ($r = .94$, $p < .0001$; Allison et al., 2012). In the AQ-10, higher scores indicate more autistic traits. This instrument was selected based on its good reported internal consistency, and high sensitivity and specificity (e.g., Allison et al., 2012). However, in this study, the internal consistency was not satisfactory ($\alpha = .43$). Therefore, given that this test has not been validated for use in Malaysia, and the reliability and validity were low, we did not use the AQ-10 diagnostically; instead, we used it to ensure that at a group level the scores were significantly different and to allow some exploration of the group-level differences in autistic traits.

#### 2.2.3.2 Repetitive Behaviours Measure

The *Repetitive Behaviour Questionnaire-2 (RBQ-2;* Leekam et al., 2007) is a 20-item parental questionnaire designed to broadly measure the severity of RRBs in both children with ASC and typical development. This evaluation tool was developed by combining two semi-structured interview behavioural measures, the Repetitive Behaviours Interview (RBI; Turner, 1996) and the Diagnostic Interview for Social and Communication Disorder (DISCO;

Wing et al., 2002). The former measure was examined, and unambiguous items were selected to form a new measure, the 33-item Repetitive Behaviours Questionnaire (RBQ; Turner, 1996). The RBQ-2 has been found to possess good psychometric properties. In the 2-year-old sample, the internal consistency of all items in the RBQ-2 was .85. The internal consistencies (Cronbach's alpha; α) for the four factors were acceptable: .80 for repetitive motor movements, .75 for rigidity/adherence to routine, .72 for preoccupation with restricted patterns of interests, and .66 for unusual sensory interest. The good internal consistency and inter-item validity suggest that the RBQ-2 is a reliable instrument for recording a range of RRBs. The total score of the RBQ-2 will be used for analyses with higher scores indicating more frequent and severe RRBs.

#### 2.2.3.3 Executive Functioning Measure

The *Childhood Executive Functioning Inventory (CHEXI;* Thorell & Nyberg, 2008) is a rating inventory for parents and teachers that consists of 24 items, measuring Executive Functioning in children between the age of 4 to 12 years. Each item is answered on a five-point scale, ranging from 1 = "definitely not true" to 5 = "definitely true" in which higher scores indicate more significant Executive Functioning challenges. This instrument is available in 21 languages, including English, Bahasa Melayu, and Chinese, and they are freely available for download on the internet (see www.chexi.se). The 24 items from CHEXI can either be divided broadly into two subscales, namely working memory (13 items) and inhibition (11 items); or four subscales: working memory (9 items), planning (4 items), inhibition (6 items) and regulation (5 items). To date all studies conducted on the CHEXI have reported that both two-factor and four-factor solutions provide a good fit for the data (Catale et al., 2015; Thorell & Nyberg, 2008). A recent confirmatory factor analysis (CFA; Camerota et al., 2018) posited that the two-factor solution is sufficient to account for the overall variance and provides the most parsimonious fit for the data, whilst the four-factor solution is redundant due to the high latent correlation between working memory and planning variables and inhibition and regulation variables. The two-factor model accounted for 41.2% of the variance in the parent rating scale and 67% of the variance in the teacher rating scale (Thorell & Nyberg, 2008). These two factors demonstrate acceptable internal consistencies (Cronbach's alpha; α): the total score was higher than .85 (Camerota et al., 2018), .89 for the Working Memory subscale and .85 for the Inhibition subscale (Catale et al., 2015). The test-retest reliability for these two subscales is also high (Catale et al., 2015): Working Memory subscale ($r = .74$, $p < .001$) and Inhibition subscale ($r = .87$, $p < .001$). The two-factor model was replicated in other studies (Camerota et al., 2018; Catale et al., 2015). All studies showed consistent results, suggesting that the CHEXI is a reliable parent- and teacher-rating instrument for measuring children's Executive Functioning. The total score of the CHEXI will be used for analyses, with higher scores representing worse Executive Functioning or greater executive deficits.

#### 2.2.3.4 Emotion Regulation Measure

The *Emotion Regulation Checklist (ERC;* Shields & Cicchetti, 1997) is a four-point rating instrument designed to assess parents' and teachers' perspectives of a child's emotion regulation ability between the age of 5 to 12 years. This measure is available in different languages, including English, Bahasa Malaysia and Mandarin. The checklist comprises 24 items, and the four response options are: never (1), sometimes (2), often (3) and almost always (4). The 24 items from the ERC are subdivided into two subscales: Emotion Regulation (8 items) and the Lability/Negativity subscale (15 items). Item 12 was not scored for either of these scales as it has not loaded on either factor in early validation analyses. The former subscale measures the appropriateness of the child's emotional expression, awareness, and empathy, with a higher score suggesting higher awareness and better regulatory processes. The latter subscale measures the child's emotional flexibility, stability, and reactivity, with a higher score suggesting lesser control over emotional intensity and expression. Confirmatory factor analyses (CFA; Kim-Spoon et al., 2013; Shields & Cicchetti, 1997) were conducted and showed acceptable internal consistency (Cronbach's alpha; α) of .89 for the total score, .83 for the Emotion Regulation subscale and .96 for the Lability/Negativity subscale. Moreover, the results have also demonstrated a high correlation between these two subscales ($r = -.50$, $p < .001$). An exploratory factor analysis (EFA; Reis et al., 2016) confirmed the two-factor solution (emotion regulation and lability/negativity) as an adequate model, accounting for 57% of the overall variance. The total score of the ERC will be used for analyses.

#### 2.2.3.5 Sensory Sensitivity Measure

In this study, we aim to focus on how sensory sensitivities affect the two-way relationship between Executive Functioning, Emotion Regulation and RRBs. The *Sensory Sensitivities Questionnaire* (Minshew & Hobson, 2008) is a 13-item instrument developed explicitly as a quick screener to measure sensory processing difficulties in ASC using lower and higher cortical sensory perception. It measures whether the participant is hyper- or hyposensitive to sensory inputs in the categories of auditory, light, tactile, pain tolerance, smell and taste. The parent-report version was used in the current study. Participants responded "yes" and "no" to questions assessing high and low pain tolerance, temperature sensitivity, tactile sensitivities, auditory and light sensitivity, awareness of smell or taste and sensitivity to environmental events. Their responses were given a score of 1 or 0 respectively, and the total score was calculated with the sum of the items, with higher scores indicating higher levels of sensory sensitivity. This questionnaire was translated into

Dutch and used in the (Lever & Geurts, 2013) study comprising a sample of 63 adults with ASC (age $M$ = 47.4 years) and 64 matched controls (age $M$ = 43.1 years). Based on that study, the total score has acceptable to good reliability of Cronbach's alpha; $\alpha$= .77. The total score of the SSQ will be used for analyses.

#### 2.2.3.6 Social Pragmatic Measure

The *Pragmatic Abilities Questionnaire (PAQ;* Jafari et al., 2019) is a 40-item questionnaire used to measure the social communication abilities in children with neurodevelopmental disorders, specifically for children with ASC and social (pragmatic) communication disorder (SPCD). This questionnaire has been used with children between 5 to 9 years old, and all items in the questionnaire are scored using a six-point scale: 0 = "never," 1 = "very rarely," 2 = "rarely," 3 = "occasionally," 4 = "very frequently," and 5 = "always," with higher scores indicating better social pragmatic skills. The development of this evaluation tool involved three stages. In stage one, 14 experts with five years or more of professional work experience and 15 mothers of children with neurodevelopmental disorders, including ASC, SPCD, Attention-Deficit / Hyperactivity Disorder (ADHD) and Specific Language Impairment (SLI), were recruited. After analysing the interview transcripts and observational results, 119 items were identified based on the Pragmatic Protocol (Prutting & Kittchner, 1987). Ten experts with five years or more of professional work experience evaluated these items based on a five-point Likert scale, with 1 representing "very irrelevant item" and 5 representing "very relevant item" in stage two. The items were reduced from 119 to 80. The rater reliability was tested through Intraclass Correlation Coefficient (ICC), and a high inter-rater agreement about the PAQ items was reported ($r$ = .78, $p < .01$). In the final stage, 185 typically developing children and 120 children with neurodevelopmental disabilities were tested with the PAQ. This process further reduced the number of items from 80 to 40 through the Rasch Rating Scale Model (RSM) measure. Any items with a Mean Square (MnSq) above 1.4 and a Z-standardized (Zstd) above +2 were considered a poor representation and were taken out from the PAQ.  A component analysis of the PAQ was conducted based on the Rasch model. The final version of PAQ was reported to explain 63.8% of the variance and had a higher person measure reliability of .97 with a person separation index of 6. The high person reliability indicates that the PAQ can effectively measure an individual's pragmatic abilities based on their estimated level. The item measure reliability was .99, with an item separation index of 10.9, indicating that the items in the PAQ have different levels of difficulty and can potentially be grouped into ten categories. The PAQ seems to be a reliable instrument to measure the pragmatic social skills of ASC, given its high standard of psychometric properties. The total score of the PAQ will be used for analyses.

**2.2.3.7 Cognitive Ability Measure**

The *Waschbusch's Parent-Rated General Cognitive Ability Scale* (Waschbusch et al., 2000) is a 20-item evaluation instrument originally designed to explore whether parents or caregivers can adequately estimate their child's general cognitive ability. To our knowledge this is, thus far, the only rating scale used to measure general intelligence completed by parents or caregivers. This measure was developed based on Cattell-Horn's theoretical models of intelligence, a theory suggesting multiple cognitive abilities nested under general intelligence to explain an individual's cognitive functioning. The 20 items were categorised into five subscales: fluid reasoning, comprehension knowledge, visual processing, auditory processing, and acquisition and retrieval of information. Four items in each subscale were identified based on the five cognitive abilities from the Cattell-Horn theory, which are nested under the broad cognitive functioning. The sixth subscale was the general cognitive ability scale, a summary score of all 20 items. The items from this questionnaire were scored using a four-point Likert scale, with 0 indicating “not at all” and 3 indicating “very much.” In the initial study, 145 children between the age of 5 to 12 years completed the Woodcock-Johnson Psycho-Educational Battery—Revised (WJ-R) and Wechsler Intelligence Scale for Children—Third Edition (WISC-III). At the same time, their parents were also given the *Waschbusch’s Parent-Rated* General Cognitive Ability Scale (Waschbusch et al., 2000). The performance reported by the General Cognitive Ability scale was significantly correlated with the WJ-R (Pearson correlation, $r = .49$, $p < .001$ for boys and $r = .51$, $p < .001$ for girls) and the WISC-III ($r = .51$, $p < .001$ for boys and $r = .43$, $p < .01$ for girls). Acceptable internal consistencies (Cronbach’s alpha; $\alpha$) were also reported in the study: .60 for the fluid reasoning scale, .66 for the comprehension-knowledge scale, .74 for the visual processing scale, .68 for the auditory processing scale, .76 for the acquisition and retrieval scale and .89 for the general cognitive ability scale. Waschbusch’s study has not only shown parents can be valid informants about their child’s cognitive abilities, these 20 items also formed the first reliable parent-rated scale that can be used to measure cognitive abilities, especially for children with difficulties completing psychoeducational assessments.

#### 2.2.4 Procedure

Once informed consent had been provided, participants were given an access link to the online questionnaires. Following completion, parents could request an interim report to understand their child’s Executive Functions, Emotion Regulation and RRBs based on the results from the questionnaires. The participants were thanked for their time with RM 10 AEON voucher.

### 2.2.5 Data Analysis

The statistical analyses were conducted using IBM SPSS Statistics for Windows, Version 25.0. Missing data were replaced with the series mean method if the data set had less than 25% missing data; otherwise, listwise deletion was used. The mean of the non-missing values of the participant in the target instrument was calculated and then replaced the missing values separately and independently from other participants within the same instrument. Shapiro-Wilk test found normality in all questionnaires except the RBQ-2, Waschbusch's Parent-Rated General Cognitive Ability Scale and PAQ. A two-step procedure: 1) transformation to uniformity and 2) transformation to normality (Templeton, 2011) were performed on these variables and the transformed data were used for analyses.

An Independent two-tailed t-test was conducted to compare all the results from the TD and ASC groups. The Pearson correlation coefficients were computed to examine the strength of the relationship between all variables. The impact of Executive Function on Emotion Regulation was also examined with linear regression.

As mentioned in the Introduction, Emotion Regulation is hypothesized to mediate the relationship between Executive Functioning and RRBs in both TD and ASC groups and this chapter aims to examine this two-way relationship using Hayes' PROCESS mediation analysis model 4 (see Figure 2.4). The bootstrapping method was used to analyse the standard errors of the data set (Hayes & Rockwood, 2020).

Monte Carlo power analysis for mediation models was used to determine the power of the study. Monte Carlo power analysis for mediation is a set of freely downloadable online applications written by Schoemann et al., (2017) in the R statistical computing language (R Core Team, 2016). A post hoc power analysis revealed that the power of the mediation effect from Executive Functioning to Emotion Regulation and then to RRBs was .80 in the total sample, .83 in the ASC population and .09 in the TD population.

Each instrument's reliability and internal consistency were checked using Tau-equivalent reliability, also known as Cronbach's alpha or coefficient alpha. An alpha value greater than .60 is acceptable (Griethuijsen et al., 2015), .45 to .55 is not satisfactory, and any value below is low or unacceptable (Taber, 2018). The internal consistency was found acceptable for most of the questionnaires used in this study: RBQ-2 (Cronbach's alpha; α = .85), CHEXI (α = .97), ERC (α = .87), Waschbusch's Parent-Rated General Cognitive Ability Scale (α = .96), and PAQ (α = .98). The internal consistency was low for the AQ-10 (α = .43) and not satisfactory for the SSQ (α = .57).

## 2.3 Results

### 2.3.1 RRBs across ASC and TD groups

The means and SDs for the RBQ-2, CHEXI, ERC, SSQ, Waschbusch's General Cognitive Ability Scale and PAQ across the ASC and TD groups are presented in Table 2.2.

**Table 2.2.**

*Descriptive statistics of the study variables*

| Domains | Subdomains and Total | *M (SD)* | |
|---|---|---|---|
| | | TD (*n* = 31) | ASC (*n* = 27) |
| Age | | 8.23 (3.05) | 7.85 (2.76) |
| Autism Traits | AQ-10 Total Score | 3.29 (1.37) | 5.81 (1.69) *** |
| Restricted and Repetitive Behaviours | RBQ-2 Repetitive Sensory/Motor | 13.24 (3.14) | 18.32 (3.85) *** |

| | | | |
|---|---|---|---|
| | RBQ-2 Insistence on Sameness | 12.84 (2.72) | 17.59 (3.48) *** |
| | RBQ-2 Total Score | 25.20 (4.84) | 34.24 (5.55) *** |
| Executive Functioning | CHEXI Working Memory | 26.54 (10.53) | 44.54 (10.04) *** |
| | CHEXI Inhibitory Control | 28.61 (9.34) | 39.56 (7.59) *** |
| | CHEXI Total Score | 55.15 (19.37) | 84.11 (16.76) *** |
| Emotion Regulation | ERC Lability/Negativity | 27.81 (7.18) | 33.47 (5.84) ** |
| | ERC Emotion Regulation | 26.16 (2.51) | 23.04 (3.32) *** |
| | ERC Total Score | 73.36 (8.29) | 64.57 (8.06) *** |
| Sensory Behaviours | SSQ Total Score | 15.26 (2.14) | 17.49 (3.18) *** |
| Intelligence Quotient (IQ) | Waschbusch's Cognitive Ability Scale Total Score | 10.07 (11.89) | 32.75 (12.71) *** |
| Social Pragmatic | PAQ Total Score | 186.46 (34.74) | 119.84 (39.92) *** |

*Note.* **p* < .05, ***p* < .01, ****p* < .001

For autistic traits, parents with TD children reported significantly lower mean scores ($M$ = 3.29, $SD$ = 1.37) than ASC children ($M$ = 5.81, $SD$ = 1.69, $t$(50.11) = -6.19, $p$ < .001). For the RRBs domain, parents indicated TD children had significantly fewer RRBs in the RBQ-2 ($M$ = 25.20, $SD$ = 4.84) than ASC children ($M$ = 34.24, $SD$ = 5.55; $t$(50.11) = -6.49, $p$ < .001). TD children were reported to have significantly lower and therefore better Executive

Functioning skills as measured by the CHEXI ($M$ = 55.15, $SD$ = 19.37) than ASC children ($M$ = 84.11, $SD$ = 16.76; $t(56.00) = -6.10$, $p < .001$). Parents also reported that TD children had significantly better emotion regulation ability measured by the ERC ($M$ = 73.36, $SD$ = 8.29) than ASC children ($M$ = 64.57, $SD$ = 8.06; $t(55.29) = 4.09$, $p < .001$).

A Pearson's correlation was conducted to examine the relationship Executive Functioning, Emotion Regulation and RRBs, and the results are shown in Table 2.3.

**Table 2.3.**

*Pearson's Correlations Table for Total scores of the AQ-10, RBQ-2, CHEXI and ERC.*

| | 1. AQ-10 | | | 2. RBQ-2 | | | 3. CHEXI | | | 4. ERC | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | TD | ASC | Total | TD | ASC | Total | TD | ASC | Total | TD | ASC | Total |
| 1. AQ-10 | - | - | - | | | | | | | | | |
| 2. RBQ-2 | .44* | .16 | .59*** | - | - | - | | | | | | |
| 3. CHEXI | .22 | .53** | .62*** | .47** | .38 | .66*** | - | - | - | | | |
| 4. ERC | -.43* | -.47* | -.61*** | -.37* | -.62*** | -.62*** | -.62*** | -.58** | -.72*** | - | - | - |
| 5. SSQ | -.06 | -.03 | .22 | .25 | .47* | .51*** | .15 | .18 | .36** | -.27 | -.41* | -.46*** |
| 6. Waschbusch's Scale | .16 | .49* | .63*** | .42* | .16 | .61*** | .75*** | .76*** | .85*** | -.36* | -.53* | -.61*** |
| 7. PAQ | -.50* | -.23 | -.63*** | -.23 | -.26 | -.58*** | -.37* | -.51* | -.67*** | .11 | .39 | .48*** |

*Note.* *p < .05, **p < .01, ***p < .001

Abbreviations: AQ-10, Autism Spectrum Quotient - 10 items, CHEXI, Childhood Executive Functioning Inventory, ERC, Emotion Regulation Checklist, PAQ, Pragmatic Abilities Questionnaire, RBQ-2, Repetitive Behaviours Questionnaire-2, SSQ, Sensory Sensitivity Questionnaire, Waschbusch's Scale, Waschbusch's Parent-Rated General Cognitive Ability Scale.

The relationship between the total scores of RBQ-2, CHEXI, and ERC were correlated in the total sample. A significant correlation was found between CHEXI and ERC in the total sample (r = -.59, *p* < .001), ASD group (r = -.58, *p* = .001) and TD group (r = -.62, *p* < .001). A significant correlation was found between CHEXI and RBQ-2 in the total sample (r = .66, *p* < .001) and TD group (r = .47, *p* = .01) but not in the ASC group (r = .47, *p* = .06). A significant correlation was found between ERC and RBQ-2 in the total sample (r = -.62, *p* < .001), TD group (r = -.37, *p* = .04) and ASC group (r = -.62, *p* < .001).

The SSQ significantly correlated with the RBQ-2 in the total sample (r=.51, p < .001) and in the ASC group (r=.47, p = .01). The relationship between the Waschbusch's General Cognitive Ability Scale was significantly correlated with all instruments besides AQ-10 score in TD group and RBQ-2 in ASC group. The relationship between the Waschbusch's Cognitive Ability Scale and the CHEXI is significantly correlated in the TD group (r= .75, *p* < .001), ASC group (r = .76, *p* < .001) and the total sample (r = .85, *p* < .001). The PAQ was significantly correlated with the AQ-10 in TD group (r= -.50, *p* = .02) and total sample (r= -.63, *p* < .001), CHEXI in TD group (r= -.37, *p* = .01), ASC group (r= -.51, *p* = .01), and total sample (r= -.67, *p* < .001), and RBQ-2 (r= -.58, *p* < .001) and ERC (r= .48, *p* < .001) but only in the total sample .

### 2.3.2 The relationship between Executive Functioning and Emotion Regulation

As can be seen from Table 2.3., the CHEXI is strongly and negatively correlated with the ERC in the total sample (r = -.59, *p* < .001), ASD group (r = -.58, *p* = .001) and TD group (r = -.62, *p* < .001), indicating that better Executive Functions correlate significantly with better emotion regulation. A partial correlation was conducted to determine the relationship between the CHEXI and ERC whilst controlling for IQ (measured with the Waschbusch's Cognitive Ability Scale). There was a strong partial correlation between the CHEXI (68.63 ±

23.19) and ERC (69.27 ± 9.24) in the total sample whilst controlling for the Waschbusch's score (20.35 ± 17.77), which is statistically significant, r(55) = -.50, $p < .001$. A strong significant partial correlation was also found between the total score of CHEXI (68.63 ± 23.19) and ERC (69.27 ± 9.24) in the TD group whilst controlling for Waschbusch's score (8.45 ± 8.92) , r(28) = -.55, $p = .002$. The relationship between CHEXI (84.10 ± 16.76) and ERC (64.57 ± 8.06) was only moderately correlated, r(24) = -.44, $p = .026$, whilst controlling for Waschbusch's score (34.00 ± 15.44). This finding indicated that IQ (Waschbusch's score) had very little influence in controlling for the relationship between Executive Functions (measured with CHEXI) and Emotion Regulation (measured with ERC) in the ASC group.

Simple linear regression was used to test if CHEXI total score significantly predicted ERC total score. In the total sample, the fitted regression model was: ERC total score = 88.81 + (-.28)*(CHEXI total score). The overall regression was statistically significant ($R^2 = .51$, $F(1, 56) = 58.50$, $p < .001$). It was found that CHEXI total score significantly related to ERC total score ($\beta = -.28$, $p < .001$). In the TD group, the fitted regression model was: ERC total score = 88.09 + (-.27)*(CHEXI total score). The overall regression was statistically significant ($R^2 = .39$, $F(1, 29) = 18.53$, $p < .001$). It was found that CHEXI total score significantly predicted ERC total score ($\beta = -.27$, $p < .001$). In the ASC group, the fitted regression model was: ERC total score = 88.19 + (-.28)*(CHEXI total score). The overall regression was statistically significant ($R^2 = .34$, $F(1, 25) = 12.92$, $p = .001$). It was found that CHEXI total score significantly related to ERC total score ($\beta = -.28$, $p = .001$). The regression table for the relationship between Executive Functioning and Emotion Regulation is presented in Table 2.4.

**Table 2.4.**

*Regression Analysis Summary for Executive Functioning (EF) Relating to Emotion Regulation (ER) in the total sample.*

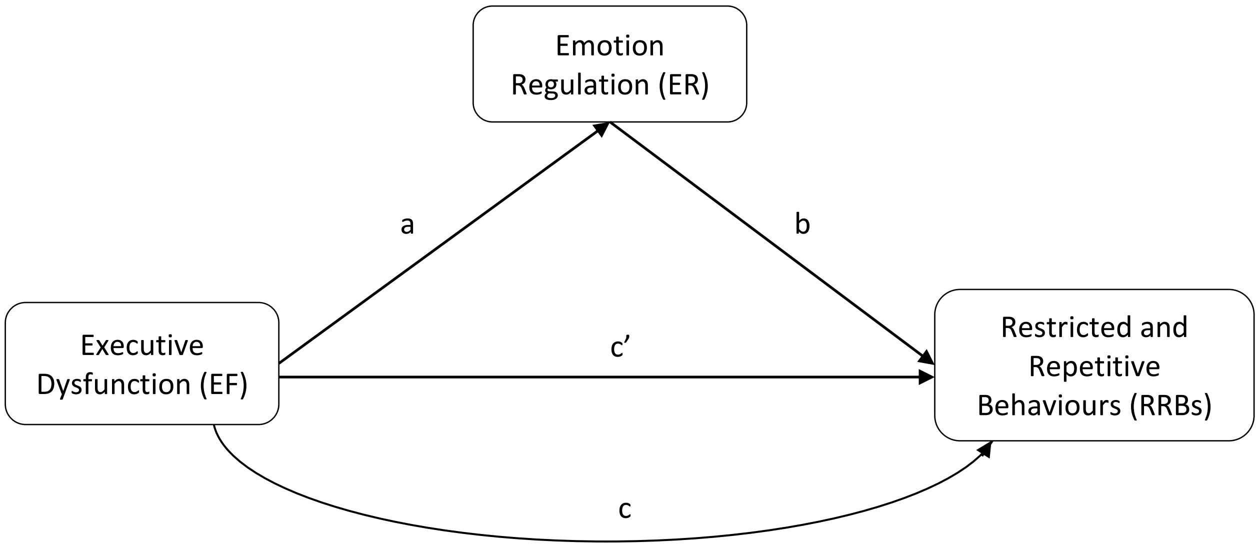
| Variable | | B | 95% CI | β | t | p |
|---|---|---|---|---|---|---|
| Total Sample | (Constant) | 88.81 | [83.41, 94.20] | | 32.96 | <.001 |
| | Executive Functioning (CHEXI) | -.29 | [-.36, -.21] | -.72 | -7.65 | <.001 |
| TD | (Constant) | 88.09 | [80.68, 95.49] | | 24.33 | <.001 |
| | Executive Functioning (CHEXI Total Score) | -.27 | [-.39, -.14] | -.62 | -4.30 | <.001 |
| ASC | (Constant) | 88.19 | [74.40, 101.98] | | 13.17 | <.001 |
| | Executive Functioning (CHEXI Total Score) | -.28 | [-.44, -.12] | -.58 | -3.60 | .001 |

### 2.3.3 Emotion Regulation as Mediator

The roles of Emotion Regulation as a mediator in the relationship between Executive Functioning and RRBs were investigated using Hayes' PROCESS model 4. The proposed relationship between Executive Functioning, Emotion Regulation and RRBs are presented in Figure 2.4.

**Figure 2.4.**

*The proposed relationship between EF, ER and RRBs in Hayes' PROCESS model 4.*




The mediating effect of Emotion Regulation (ERC total score) on the relationship between Executive Functioning (CHEXI total score) and RRBs (RBQ total score) is presented in Table 2.5.

**Table 2.5.**

*Mediating effect of Emotion Regulation in the relationship between Executive Functioning and RRBs.*

| | TD (*n* = 31) | ASD (*n* = 27) | Total (*n* = 58) |
|---|---|---|---|
| a | -.267* | -.281* | -.285* |
| b | -.052 | -.527* | -.298* |
| Direct effect, c' | .068 | .046 | .127* |
| Total effect, c | .081* | .194* | .212* |
| Indirect effect, a*b | .014 | .148* | .085* |
| 95% CI | [-.024, .068] | [.035, .337] | [.023, .165] |

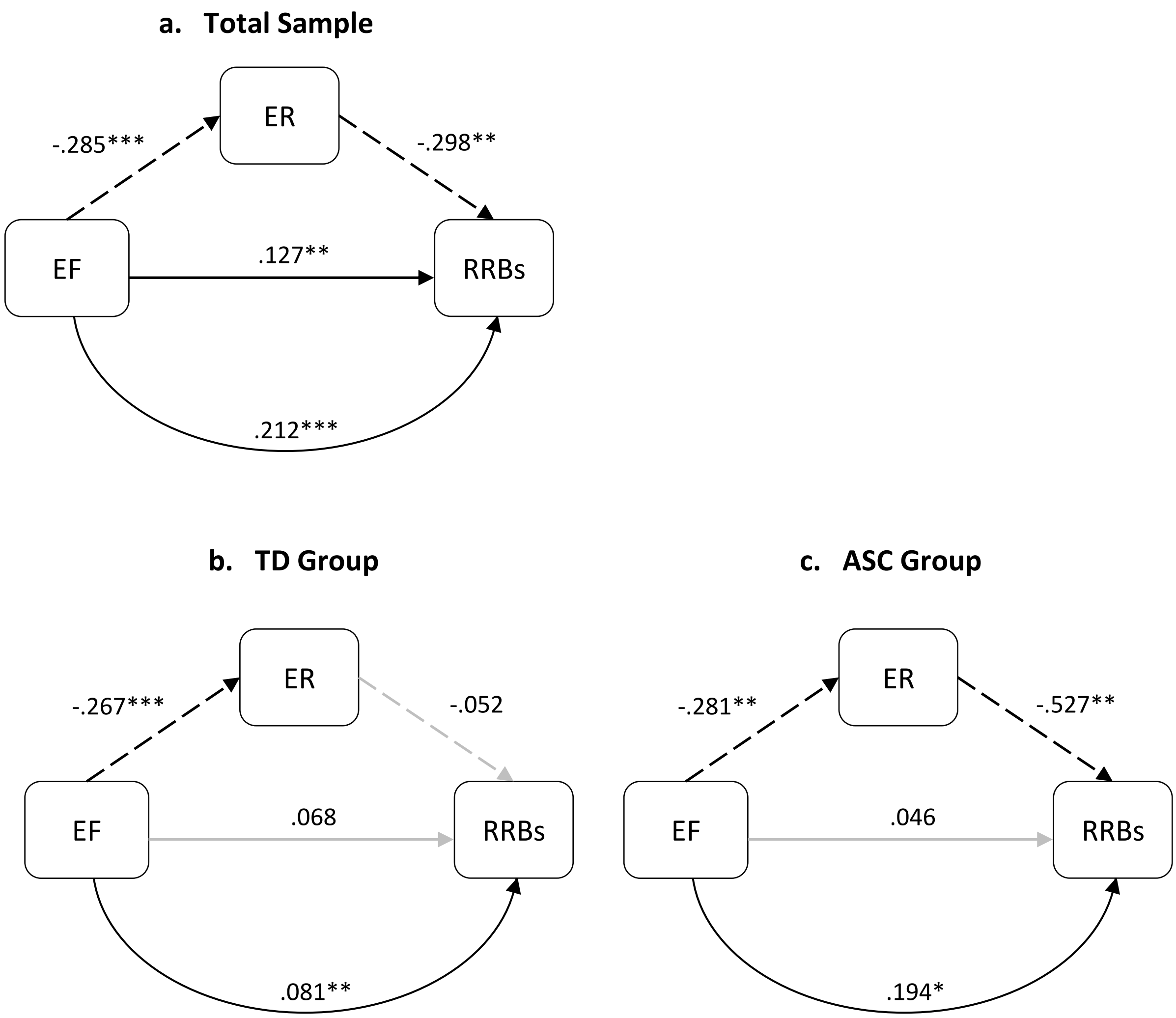
*Note:* The significance of the effects was assessed through the 95% bootstrap confidence interval (CI). The asterisk*, represents a significant result, i.e., the 95% bootstrap CI does not contain zero. The total effect, c, is the sum of direct effect, c', and indirect effect, a*b.

In the total sample and ASC group, a significant indirect effect, a*b, of Executive Functioning to Emotion Regulation to RRBs, was found. A significant direct effect, c', of Executive Functioning on RRBs was also observed. The total effect, c, is the sum of direct and indirect effects, a*b + c', and it was significant in this sample. There was evidence indicating a significant effect of Executive Functioning on Emotion Regulation and Emotion Regulation on RRBs. Interestingly, Emotion Regulation showed a mediating effect on the relationship between Executive Functioning and RRBs in the ASC sample.

In the TD sample, the indirect effect, a*b, and the direct effect, c', were not significant. The total effect, c, was significant. A significant effect of Executive Functioning on Emotion Regulation, a, was found. The effect of Emotion Regulation on RRBs, b, was not significant. A mediation diagram illustrating the relationship between the study variables is presented in Figure 2.5.

**Figure 2.5.**

*A mediation diagram illustrating the relationship between the study variables.*

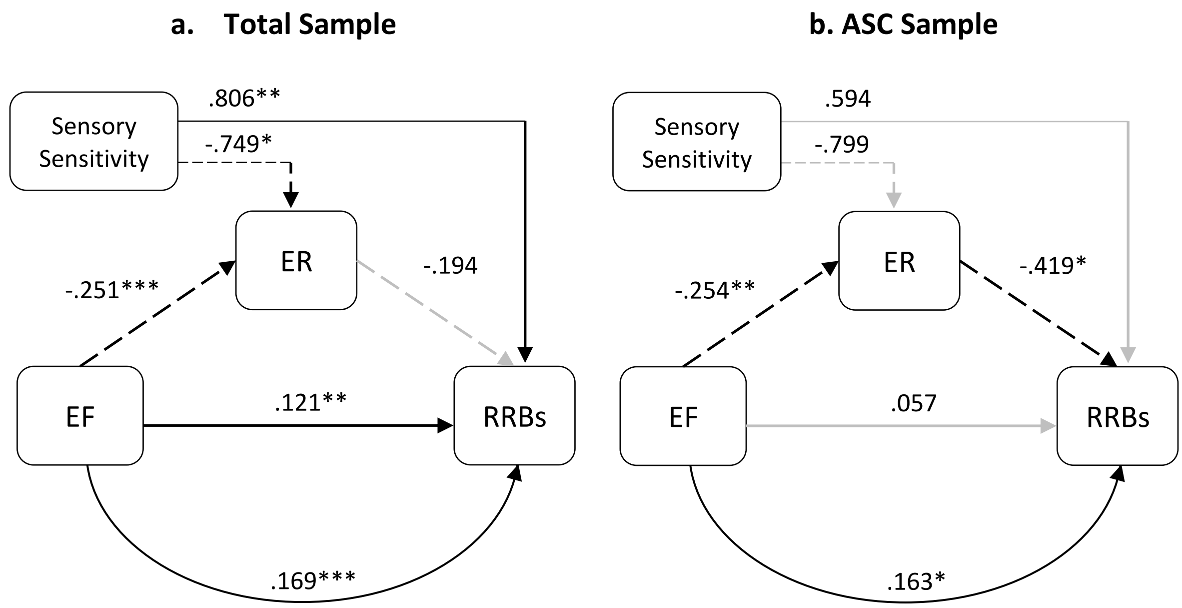



*Note:* Solid lines refer to positive values and dotted lines refer to negative values; black lines refer to significant results and grey lines refer to non-significant results.
*Note.* *p < .05, **p < .01, ***p < .001

A Pearson's Correlation analysis reported that the relationship between sensory sensitivity and Executive Functions (r = .36, *p* =.006), Emotion Regulation (r = -.46, *p* <.001) and RRBs (r = .57, $p$ < .001) were highly significant in the total sample. As reported earlier, Emotion Regulation both serve as a mediator in the relationship between Executive Functioning and RRBs in the total sample and the ASC group; therefore, for exploratory purposes, this relationship was further analysed with sensory sensitivity (measured with SSQ) as a covariate in these two datasets (the total sample and ASC group), as presented in a diagram in Figure 2.6. In the total sample, the relationship between Emotion Regulation and RRBs was no longer significant whilst controlling for sensory sensitivity whereas Emotion Regulation remained a mediator in the ASC group.

**Figure 2.6.**

*Emotion Regulation as a mediator in the relationship between Executive Functioning and RRBs in the total sample and the ASC group with sensory sensitivity as a covariate.*




*Note:* Solid lines refer to positive values and dotted lines refer to negative values; coloured lines refer to significant results and grey lines refer to non-significant results.

*Note.* *p < .05, **p < .01, ***p < .001

## 2.4 Discussion

### 2.4.1 Autistic Traits and RRBs across the ASC and TD Groups

The first aim of this chapter was to explore the differences in autistic traits, RRBs, Executive Functioning and Emotion Regulation across a sample of 31 TD children and 27 children with ASC between 4 to 13 years old with parent-report questionnaires. It is important to note that this study was conducted during the Covid-19 pandemic. While acknowledging the limitations of using only questionnaires to answer the research questions compared to, arguably, more objective neuropsychological testing and behavioural observation, questionnaires do provide higher flexibility in implementation, allowing scientific progression even whilst social restrictions were in place.

Almost all the questionnaire scores between the ASC group and the TD group were significantly different. As expected, the ASC group had significantly higher autistic trait scores than the TD group, though we note the average score only approached the cut-off value of 6 on the AQ-10, despite previous research showing Malaysians scored higher in the AQ than other populations (Chee et al., 2023). Additionally, the ASC group parents reported significantly more RRBs than the parents in the TD group. Pearson's correlations confirmed that children with more reported autistic traits had higher levels of reported RRBs.

### 2.4.2 Executive Functioning and Emotion Regulation

The second aim of this chapter is to examine the relationship between Executive Functions and Emotion Regulation. In line with Sudikoff et al. (2015) and Jahromi et al. (2013), we found better Executive Functions are correlated with better abilities in regulating their emotions in both TD and ASC groups. This relationship remained significant even when the influence of IQ was controlled.

Typically developing children were reported to have better Executive Functions and manage their emotions better than children with ASC. When the relationship between Executive Functions and Emotion Regulation was further tested with regression, a positive relationship was found.

### 2.4.3 Mediator between Executive Functioning and RRBs

The third aim of this chapter was to provide a novel exploration into the role of Emotion Regulation in the relationship between Executive Functioning and RRBs. Previous research on RRBs focused mainly on one specific characteristic, atypical sensory sensitivities. Before the publication of the DSM-5, atypical sensory sensitivity was commonly related to autism but not inevitable. Then in the DSM-5, atypical sensory sensitivity was included under the category of RRBs. Thus, rather than examining the conventional relationship between sensory sensitivity and RRBs, this study examined Executive Functioning and its relationship with RRBs.

First, we examined the relationship between RRBs, Executive Functioning and Emotion Regulation. All relationships were significant in the total sample [as presented in Figure 2.5 (a)]. The direct effect, c', from Executive Functioning to RRBs were not significant in both TD and ASC groups. Besides the possible sampling bias, the non-significant relationship might also indicate that the relationship between Executive Functioning and RRBs might be mediated or moderated by other variables. Then, we examined the mediation path and found a significant path from Executive Functioning → Emotion Regulation → RRBs, indicating Emotion Regulation was a significant mediator of the relationship between Executive Functioning and RRBs, in the ASC group but not in the TD group. However, this result should be approached with caution due to its low power (.10) in the TD group. Future investigation should be undertaken with a higher number of participants to examine whether Emotion Regulation is also the mediator between Executive Functioning and RRBs in the TD group.

As mentioned previously, sensory sensitivity has frequently been related to occurrences of RRBs, and thus, the relationship (Executive Functioning – Emotion Regulation – RRBs) was further examined in the total sample and the ASC group whilst including sensory sensitivity as a covariate. This investigation was only conducted in the total sample and the ASC group as Emotion Regulation was found to mediate the relationship between Executive Functioning and RRBs in these two analyses, and the power of this relationship was acceptable (total sample = .83; ASC group = .88). A significant direct effect was found between sensory sensitivity and both Emotion Regulation and RRBs in the total sample, but no significant relationship was found between sensory sensitivity and Emotion Regulation nor RRBs in the ASC group. The relationship between sensory sensitivities and Emotion Regulation in the ASC group did not support past studies that atypical sensory sensitivities are likely to result in emotions such as anxiety (Black et al., 2017; Green et al., 2012).

The direct relationship between sensory sensitivity and RRBs was significant in the total sample, which supports previous findings that sensory sensitivity and RRBs are closely related. Surprisingly, the direct relationship between sensory sensitivity and RRBs was not significant in the ASC group despite that it has been categorised under the same main characteristic in the DSM-5. It is critical to consider the low reliability ($\alpha$ = .57) of the SSQ that was used to measure the sensory sensitivity while interpreting this result. It is postulated that the type of sensory domain can be related to different behavioural traits. For example, Schulz & Stevenson (2020) reported that typically developing adults with greater visual sensitivity are more likely to display insistence on sameness and show less interest in social interactions. The SSQ, which focuses on only four main domains: low temperature/pain tolerance, high temperature/pain tolerance, tactile and overall sensory sensitivities may not capture other sensory domains such as visual, auditory, vestibular, proprioception and olfactory domains. This limitation may affect the relationship between sensory sensitivity and RRBs considering individuals with ASC have been reported to experience various of sensory problems (Robertson & Baron-Cohen, 2017). Besides that, as mentioned earlier, sensory sensitivity has been included in RRBs in the DSM-5 and therefore, when being analysed again as an independent construct, the specific sensory sensitivity did not have a significant effect on the Emotion Regulation and RRBs. As a result, the Emotion Regulation remained as the mediator in the relationship between Executive Functioning and RRBs in the ASC group.

The executive dysfunction hypothesis has been used to explain the behaviours of individuals with autism (Demetriou et al., 2018; Turner, 1997). Studies found that poorer Executive Functions, such as cognitive flexibility, response inhibition, and working memory, are associated with and predict a higher level of RRBs (Faja & Nelson Darling, 2019; LeMonda et al., 2012; Lopez et al., 2005; Reed et al., 2013). The empirical findings are mixed (Jones et al., 2018), and the developmental trajectory account suggested focusing on the development of cortico-striatal-circuits and neurobiological changes alongside the occurrence of RRBs (Leekam et al., 2011). It is plausible that these mixed findings were due to an overlooked mediator between Executive Functions and RRBs. The findings from the current study confirmed the involvement of Emotion Regulation as an additional mediator in the relationship between Executive Functioning and RRBs.

#### 2.4.4 Limitations

There are two major limitations in this chapter. The first limitation in this Chapter is the small sample size consisting of 31 typically developing children and 27 children with ASC.

As one of the main aims of this chapter is to examine the role of Emotion Regulation in the relationship between Executive Functioning and RRBs using the mediation model, the power of the mediation effect was examined with Monte Carlo power analysis for mediation model 4. The analysis found a moderate power of .83 for the total sample, .88 for the ASD group and low power of .10 for the TD group. The interpretation of the results could therefore be constrained by the small sample size, especially for the TD group.

The second limitation of this chapter is that only parent-report questionnaires were employed to measure Executive Functioning, Emotion Regulation and RRBs. Due to practical constraints, we did not have the opportunities to use arguably more objective measures such as performance-based and observational-based measures. Firstly, although the RBQ-2 allowed parents to report the severity of their child's RRBs based on their general observation, the questionnaire failed to capture the wide-ranging topographies and variety of repetitive sensorimotor behaviours and their occurrences from aspects such as frequency, duration, and amplitude. The results can also be affected by parental bias. Grahame et al. (2015) argued that the total score of the RBQ-2 is not sensitive to capturing changes in RRBs, and the previous development work had identified the importance of documenting the levels of and any changes in RRBs across different social-emotional contexts. Therefore, using the RBQ-2 alone may not be enough in examining participants' RRBs. Secondly, previous studies have shown that neuropsychological (or performance-based) testing and ratings of Executive Functioning are often not strongly correlated (Mcauley et al., 2010; Thorell et al., 2010). These findings suggest that results from neuropsychological tests and rating scales might capture different aspects of Executive Functions. Using both neuropsychological tests and rating scales can provide more comprehensive and in-depth information regarding a specific aspect of Executive Functioning. Based on the above limitations, changes were made to the next study, reported in Chapter 3, to replicate and extend the current study.

### 2.4.5 Summary

This chapter has provided an exciting preliminary exploration of the effect of Executive Functioning and Emotion Regulation on RRBs in children with ASC. Firstly, this chapter has further confirmed that children with autism have higher levels of RRBs and autistic traits and have significantly poorer Executive Functioning and Emotion Regulation than typically developing children. Consistent with previous studies, Executive Functioning was associated with Emotion Regulation in both TD and ASC children. Finally, we found Emotion Regulation serve as a mediator in the total sample and ASC group but not the TD group.

# 3 Chapter Three: Restricted and Repetitive Behaviours (RRBs) across three Emotional Contexts

## 3.1 Introduction

The existing research has consistently shown that individuals with Autism Spectrum Conditions (ASC) who have weaker Executive Functioning usually have more Restricted and Repetitive Behaviours (RRBs, Iversen & Lewis, 2021). Studies have also found that RRBs may serve as an Emotion Regulation mechanism (Rodgers et al., 2012) and higher levels of RRBs have been observed in children with ASC during emotional contexts, especially in those with lower cognitive abilities (Willemsen-Swinkels et al., 1998). These findings suggest potential roles of Executive Functioning and Emotion Regulation in RRBs. Interestingly, Executive Functioning has also been found to be related to Emotion Regulation. Children's performance on Executive Functioning tasks was significantly related to their ability in regulating emotions (Carlson & Wang, 2007) and Executive Functioning training improved children's emotional competence (Li et al., 2020). Despite observed relationships between Executive Functioning and RRBs, Emotion Regulation with RRBs, and Executive Functioning and Emotion Regulation, no studies have looked at the relationship between Executive Functioning, Emotion Regulation and RRBs altogether in autism.

In Chapter 2, we examined this two-way relationship and found that Emotion Regulation played a mediating role in the relationship between Executive Functioning and RRBs in the total sample and the ASC group, but not in the typically developing (TD) group. Emotion Regulation remained a mediator in the ASC group whilst controlling for sensory sensitivity. In the total sample, the relationship between sensory sensitivity and RRBs was strongly significant, suggesting that sensory sensitivity could be one of the direct underlying mechanisms of RRBs. In the ASC group, the relationship between sensory sensitivity and RRBs was not significant. This finding suggests that any relationship between sensory sensitivity and RRBs can be mediated by other variables. However, it is important to note that, as this study was conducted during the Covid-19 pandemic, we relied on subjective parent-report measures. The use of only questionnaire data is not ideal in terms of identifying the underlying neurophysiological mechanisms and potential causal relationships of the target constructs due to the limitations of their design.

Therefore, in order to experimentally manipulate the relationship between Executive Functioning, Emotion Regulation and RRBs, we recorded RRBs across different emotional contexts. Then, we replicated our previous study to investigate the overall robustness of the patterns found in Chapter 2, and extend the previous study by using neuropsychological tasks and observational measures to examine whether Emotion Regulation remained as a mediator in the relationship between Executive Functioning and RRBs when different measures are used.

### 3.1.1 Manipulation of Emotional Contexts and RRBs

As mentioned in Chapter 1, typically developing children and children with developmental delay (e.g., autism, attention deficit/hyperactivity disorder, developmental language disorder) with high intelligent quotient (IQ) above 80 displayed more RRBs during "composure" emotion, an emotional state when they were not showing any signs of excitement (Willemsen-Swinkels et al., 1998). Children with moderate IQ between 50 and 80 displayed more RRBs when they were excited and "composed", and children with low IQ below 50 displayed RRBs across all three conditions: composure, excitement, and distress. Their emotions were determined based on their facial expressions and the content of their verbal and nonverbal communication. Based on these findings, children with high IQ seem to regulate their emotions with other strategies during excitement and distress while children with lower IQ used RRBs to regulate their emotions when they were experiencing different emotions. RRBs have also been posited to serve different functions across different emotional situations (Willemsen-Swinkels et al., 1998). During distress, RRBs were considered an outlet that helped to soothe external stimulation by redirecting the child's attention. They also served as an expression during excitement, and a behaviour to attract others' attention during "composure" situations. Although a relationship has been found between specific emotions and RRBs in other studies (Black et al., 2017; Joosten et al., 2012; Lidstone et al., 2014; Rodgers et al., 2012), the findings from Willemsen-Swinkels et al. (1998) are arguable as the emotion valence was decided post-hoc rather than a priori based on operational definitions. The participants' emotions were neither determined by the context nor the planned operational definitions, but their facial expressions gathered from the experiments. The post-hoc data analyses can be an issue as they do not conform the randomization model of statistical inference and the findings can simply be coincidence.

In this Chapter, we manipulated three emotional conditions: Task, Preferred and Neutral conditions, and examined participants' RRBs across these conditions. Participants were given activities during the Task condition, videos that they like during the Preferred condition and videos that they neither liked nor disliked during the Neutral condition. Based on previous studies, we predicted that participants with lower Emotion Regulation ability were more likely to exhibit RRBs to regulate emotions in the presence of intense emotions, in other words the Task and Preferred conditions compared to the Neutral. Additionally, their Emotion Regulation was predicted to be related to their Executive Functioning skills.

In addition to manipulating the emotional context, performance-based (or neuropsychological) measures were used to examine Executive Functioning, and observational-based method were used for measuring repetitive behaviours. Therefore, in the next section, we will explore the five most frequently used parent-rating and neuropsychological measures of Executive Functioning. Then, we will investigate the previous research using observational-based methods for measuring RRBs.

### 3.1.2 Measurements of Executive Functioning

Research in Executive Functioning has often been criticised for lacking reliable and valid measures, mainly due to the lack of task purity in the instruments used to measure Executive Functions (Chan et al., 2008; Nyongesa et al., 2019). At present, there are no standardized measures of Executive Functioning, but a great number of rating scales and performance-based (or neuropsychological) measures have been used to capture the broad range of underlying constructs in Executive Functioning. To systematically identify measures used in the current study, we searched for articles published in the last five years using the PubMed database with the following terms: "autism" and "executive function" and "rating" or "neuropsychological." One hundred and forty-one articles were identified and screened, and the most frequently used rating and neuropsychological/performance-based measures of Executive Functioning are described in Table 3.1. This table provides information regarding the measures which enables us to explore the suitability of each measure for this study which involves children with ASC. Here we focus on measuring tools that are solely devoted to the assessment of Executive Functioning constructs. Although measuring tools such as the Behaviour Assessment System for Children, second edition (BASC-2, Reynolds & Kamphaus, 2004) contains items regarding executive control and frontal lobe functioning, they are not included in the list as their focus is not exclusively on Executive Functioning.

**Table 3.1.**

*A list of frequently used rating and neuropsychological measures of Executive Functioning and their measuring information.*

| Measuring tool | Description | Administration | Age range | ASC (norm) data? |
|---|---|---|---|---|
| Rating measures | | | | |
| Barkley Deficits in Executive Functioning Scales (BDEFS; Barkley, 2011) | A parent-report questionnaire which contains 89 items assessing Executive Functioning domains such as Self-Management to Time, Self-Organization/ Problem Solving, Self-Restraint, Self-Motivation, and Self-Regulation of Emotion. The BDEFS has high internal consistency for typical development population (Cronbach's $\alpha$ = .93-.96). | Long form: 10-15 minutes<br>Short form: 3-5 minutes | 6-17 years | No |
| Behaviour Rating Inventory of Executive Function (BRIEF; Gioia et al., 2000) | A parent-completed questionnaire composed of 86 items that measure Executive Functioning domains such as Inhibit, Shift, Emotional Control, Initiate, Working Memory, Plan/Organize, Organization of Materials, Monitor. The BRIEF has high internal consistency ($\alpha$ = .80-.98). | 10-15 minutes | 5-18 years | Yes (HFA) |

| | | | | |
|---|---|---|---|---|
| Comprehensive Executive Function Inventory (CEFI; Naglieri & Goldstein, 2014) | A parent, teacher, and/or self-report rating scales, each with 100 items for measuring Executive Functioning domains such as Attention, Emotion Regulation, Flexibility, Inhibitory Control, Initiation, Organization, Planning, Self-Monitoring and Working Memory. The BRIEF has high internal consistency between .85-.99 for parent-report, .90-.99 for teacher-report and .77-.97 for self-report. | 15 minutes | 5-18 years | Yes |
| Childhood Executive Functioning Inventory (CHEXI; Thorell & Nyberg, 2008) | A 24-item rating instrument for parents and teachers that are freely available in 18 languages. There are four different subscales indexing inhibition, regulation, working memory and planning and two latent factors were identified (i.e., working memory and inhibition) which demonstrate high internal consistency ($\alpha > .85$). | Up to 5 minutes | 4-12 years | No |
| Executive Function Index (EFI; Spinella, 2005) | A self-rating scale which contains 27 items that can be divided into five subscales, namely Impulse Control, Strategic Planning, Organization, Motivational Drive, and Empathy. The | Up to 5 minutes | 17-60 years | No |

| | | | | |
|---|---|---|---|---|
| | EFI has good internal consistency (α = .82). | | | |
| Neuropsychological (or performance-based) measures | | | | |
| Card Sorting Task (Zelazo, 2006) | A widely used measure of Executive Function, particularly set shifting, that is suitable for participants across a broad range of ages. Participants are required to sort a series of test cards, first according to one dimension (e.g., shape), and then another dimension (e.g., colour). | Standard protocol = 5 minutes | This task has been used with participants above 3 years | Yes (Dichter et al., 2010) |
| Go/No-Go Task | A two-step verification task great for measuring a domain of Executive Functioning, namely inhibition. Participants are required to respond to certain stimuli ("go" stimuli) and make no response for other stimuli ("no-go" stimuli). | Standard protocol = 5 minutes | The Go/No-Go has been used with participants above 3 years | Yes (Uzefovsky et al., 2016) |
| Stroop Colour-Word Test | A quick, easy and standardized neuropsychological test used | Standard protocol | Children's | Not a valid |

| | | | | |
|---|---|---|---|---|
| (Golden, 1978) | for assessing a domain of Executive Functioning, namely inhibition. Participants are required to indicate the colour of the word (not by its meaning). During congruent trials, the colour word and its colour are the same, whereas during incongruent trials, the colour word and its colour are not the same. | = 10-15 minutes | version = 5-14 years | test for autism (Adams & Jarrold, 2009) |
| Tower of London Test | One of the most frequently used neuropsychological task for measuring planning ability in typical development (TD) and clinical samples. Participants are required to move an entire stack of disk to another rod following simple rules. | Standard protocol = 8 minutes | 7-80 years | Yes (Unterrainer et al., 2020) |
| Trail Making Test | A neuropsychological task that provides information about visual search speed, speed of processing, scanning, mental flexibility, and Executive Functioning. Participants are required to connect 25 consecutive targets in sequential order, which is similar to a child's connect-the-dots puzzle. | Standard protocol = 15 minutes | 6-97 years | Yes (Losh et al., n.d.) |

Abbreviations: HFA, High-Functioning Autism.

In a comprehensive review of Executive Functioning in ASC, Demetriou et al. (2019) summarized the neuropsychological tests corresponding to the discrete Executive Functioning domains they measure. The Wisconsin Card Sorting Test (WCST; Heaton et al., 1993) is among the most frequently used instruments in measuring Executive Functioning. WCST measures set-shifting, which is often referred to as a lower-level form of cognitive flexibility (Dajani & Uddin, 2015), which requires participants to sort cards by either shape, colour or number. Various studies using this measure demonstrated significant differences in cognitive flexibility between individuals with ASC and typical development (Lopez et al., 2005; Tsuchiya et al., 2005; Verté et al., 2006; Willcutt et al., 2008). However, Geurts et al. (2009) argued that poor performances on the WCST can result from other cognitive deficits. The lack of purity in neuropsychological tasks, WCST in this case, raises concerns of its appropriateness in assessing Executive Functions. This is especially the case in ASC, where individuals may have other cognitive deficits which could affect their overall performance in WCST, not necessarily poor cognitive flexibility. Using a specific instrument that examines Executive Functioning at an appropriate difficulty level is important in studies involving participants with ASC. Therefore, instead of using the WCST, a simplified version of WCST called the Dimensional Change Card Sorting (DCCS), which has been used across a wide age range of children (Zelazo, 2006) will be used in this study (see Method for more details).

Demetriou et al. (2018) demonstrated a bigger effect size when studies in ASC used behavioural rating scales to measure Executive Functioning relative to neuropsychological measures. This result suggests that behavioural rating scales may be more ecologically valid and better capture Executive Functioning in ASC (Kenworthy et al., 2008). The findings from Ten Eycke & Dewey (2016) suggested that rather than selecting between neuropsychological tests and rating scales, these measures measured different constructs of executive functioning that are associated with different performances. They examined the Executive Functions, motor, attention, mathematics and reading performance of a sample of 405 children between 5 to 18 years old. Of these children, 112 were typically developing, 130 had various combinations of attention, motor and reading challenges, 79 had attention challenges, 55 had motor challenges and 16 had reading difficulties. The performance-based executive functioning tests comprised four subtests of the NEPSY-II (Korkman et al., 2007): Animal Sorting, Auditory Attention, Response Set, and Inhibition (Inhibition-Naming, Inhibition-Inhibition, and Inhibition-Switching) whereas parent-report of Executive Functions were measured with the BRIEF. The BRIEF provides an overall score and scale scores for seven constructs: working memory, response inhibition, shift/cognitive flexibility, emotional control, planning, organization of materials, initiation, and monitoring. Results showed that motor functioning was only associated with performance-based measures of Executive Functions, whereas attention was only associated with the parent-report measure. Reading and mathematics performance were associated with both parent-report and performance-based measures. These findings highlight the importance of examining Executive Functioning with parent-report and performance-based measures to provide a more comprehensive understanding of the target behaviours. The use of only parent-report questionnaire or performance-based measure is tied to clear limitations which may hinder the interpretation of results as they evaluate different constructs of Executive Functioning (Ten Eycke & Dewey, 2016; Toplak et al., 2013). Therefore, once again, we recognise the importance of using both parent-report and performance-based measures.

### 3.1.3 Observational-based measures of Repetitive Motor Movements

In addition to using both parent-report and performance-based measure for assessing Executive Functioning, we are also using both parent-report and observational-based methods for measuring RRBs. RRBs were traditionally defined as purposeless behaviours (Bodfish et al., 2000; M. Turner, 1999), but recent studies have suggested that RRBs serve apparent functions for individuals with ASC (Leekam et al., 2011; Muskett et al., 2019; Rodgers et al., 2012; Russell et al., 2019). Individuals with ASC have reported employing RRBs as a coping and adaptive mechanism that helps them to soothe and regulate their intense and uncontainable emotions such as anxiety (Joyce et al., 2017; Wigham et al., 2015) and excitement (Kapp et al., 2019), and in response to emotional triggers (Militerni et al., 2002). The common methods used to measure RRBs include rating scales, observation methods and wearable automated technology. Rating scales typically involve informants to give a global impression of the frequency, duration and severity of an individual's RRBs (M. H. Lewis & Bodfish, 1998). Although rating scales such as the Autism Diagnostic Interview-Revised (ADI-R; Le Couteur et al., 2003), Repetitive Behaviors Scale-Revised (RBS-R; Lam & Aman, 2007), and Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007) are among the most popular measurements of RRBs (Leekam et al., 2011), they generally have well-documented limitations. From a measurement perspective, rating scales can have questionable accuracy, possibly deriving from the informant's subjective impressions of the behaviours (Johnston & Pennypacker, 2009). Rating scales often fail to capture inter-individual variation in the temporal and topographical dimensions of repetitive motor movements (McEntee & Saunders, 1997; Pyles et al., 1997), and their psychometric properties vary (Rojahn et al., 2000). Researchers have used observational measurement methods as an alternative to counteract these limitations (Goldman et al., 2009; Ozonoff et al., 2008; Stronach & Wetherby, 2014; Watt et al., 2008).

Limited studies have investigated whether direct observations provide similar information regarding RRBs as rating scales, perhaps due to the time-consuming and laborious behavioural coding process of direct observations. Direct observations of RRBs have been found to predict clinician's observational rating scores. For example, Stronach & Wetherby (2014) coded the occurrences of 55 toddlers' repetitive behaviours, who were between the age of 15 to 24 months, based on an unpublished manual Repetitive Movement and Restricted Interest Scales (RMRIS; Weatherby et al., 2011). They found repetitive movements with objects at home significantly predicted RRB scores of Autism Diagnostic Observation Schedule (ADOS; Gotham et al., 2006), but similar behaviours in a clinic setting predicted social affect scores of ADOS. Ozonoff et al. (2008) also found a significant correlation between frequency of object spinning at 12 months and ADOS repetitive behaviour algorithm scores at 36 months. ADOS Restricted and Repetitive domain is a clinician-administered observational assessment with scores ranging from 0 to 3. A score of 0 indicates absence of the particular behaviour, and higher ratings from 1 to 3 indicate higher severity. There are six items in the ADOS RRBs domain, namely, Stereotyped Language, Intonation of Vocalizations, Sensory Interests, Hand and Finger Mannerisms, Complex Mannerisms and Repetitive Behaviours (Kim & Lord, 2010b). Clinicians generally takes 30 to 60 minutes to administer a series of structured and semi-structured tasks in the ADOS and present a series of opportunities for the participant to display behaviours that are relevant to the diagnosis of autism (Akshoomoff et al., 2006). Stronach & Wetherby's (2014) findings suggested that direct observation of RRBs at home can predict clinicians' observational rating scores from the ADOS.

In parent-report rating measures of RRBs, researchers are not able to determine whether the provided information is based on specific contexts or the participants' general impressions. Experiments with direct observation enable manipulation of emotional variables and information gathered can take confounding variables into consideration.

Direct measurements also provide information about the topographies of repetitive behaviours, how frequently they occur, for how long they occur and the conditions under which they do and do not occur, which are critical for understanding the nature of RRBs. Despite the complicated process that requires undivided attention to identify, observe and record the behaviours from video recordings, this methodology provides information that rating scales and interviews are unable to. On the other hand, parent-rating scales have been claimed to provide information that may relate more to real-life situations compared to observation in a standardized context (Iversen & Lewis, 2021). Therefore, once again, we believe both parent-report and observational-based measures are important to provide a thorough view of children's RRBs.

### 3.1.4 Objectives

There are three objectives in this chapter. First, we explored the duration of RRBs across three manipulated conditions: Task, Neutral and Preferred. Then, we replicated Chapter 2 and examined the occurrences of repetitive behaviours in children with typical development (TD) and ASC using questionnaires. As an extension from Chapter 2, we also used neuropsychological tasks to measure Executive Functioning and observational-based methods to measure RRBs across three different conditions (i.e., Task, Preferred, Neutral). We then explored whether the relationship between Executive Functioning, RRBs and relevant mediator Emotion Regulation followed the same pattern as those in Chapter 2 with different participants. Finally, we explored whether using different, arguably more objective, measures lead to similar or different patterns of relationships.

As described previously in the Introduction, we expected to observe more RRBs during emotionally triggered conditions, such as Task and Preferred conditions, compared to the Neutral condition, if RRBs serve as an Emotion Regulation mechanism. Replicating

Chapter 2, we expected to observe similar patterns of findings that Emotion Regulation serves as the mediator in the relationship between Executive Functioning and RRBs when rating scales are employed. Given past studies, we hypothesized differences in patterns when rating scales are replaced with neuropsychological and observational-based methods. Finally, we hypothesized that neuropsychological tasks and rating scales of Executive Functioning are likely to correlate, as CHEXI also examines the subtypes of Executive Functioning. We believe that using both parent-report and direct observation provides comprehensive information about the children's RRBs while the observation-based methods enable us to manipulate the emotional variables during direct observation, which is crucial especially when Emotion Regulation is considered to have a mediating effect on RRBs.

## 3.2 Method

### 3.2.1 Participants

#### 3.2.1.1 Characteristics of the Participants

The sample for this study comprised 114 children, which comprised 60 typically developing (TD) children and their parents, aged 5 years to 13 years (M = 9 years 9 months, SD = 1 year 10 months), and 54 children with ASC and their parents, aged 6 years to 13 years (M = 10 years, SD = 1 year 10 months). All the children with ASC had a clinical diagnosis.

Diagnosis: Similar to Chapter 2, each child from the ASC group had a clinical diagnosis by a professional such as medical practitioner, paediatrician, family medicine specialist, or psychiatrist. These professionals are recommended diagnosis providers by the Ministry of Health Malaysia in the Clinical Practice Guidelines for Management of Autism Spectrum Disorder in Children and Adolescents (Malaysian Health Technology Assessment Section, 2014). Again, we did not use the international assessments to reassess the children's diagnosis as these assessments have not been validated in in Malaysia. Instead of the autism-spectrum quotient (AQ-10; Allison et al., 2012) that was used in Chapter 2, we used the Autism Quotient -Short (AQ-Short; Hoekstra et al., 2011) to examine the difference between TD group and ASC group.

It is important to note that one of the administrators of mainstream classes was confused with the criteria for typically developing (TD) children and recruited students who were attending remedial classes for additional academic support in subjects such as English, Bahasa Malaysia (BM), Mandarin and Mathematics. Additional data analysis for these 19 children was conducted for exploratory purposes.

#### 3.2.1.2 Ethics

This study has been reviewed by the University of Reading Malaysia Research Ethics Committee and has been given a favourable opinion for conduct with *UoRM REC 2021/03* as the unique approval reference number. All researchers working on this project have had the appropriate criminal records checks by the Ministry of Foreign Affairs and received a Certificate of Good Conduct.

#### 3.2.1.3 Recruitment Procedures

Families were recruited through public primary schools with an integrated special education programme, namely Program Pendidikan Khas Integrasi (PPKI), in Bahasa Malaysia. Approval was obtained from the Ministry of Education (MoE) Malaysia and the Department of State Education (JPN). Following their approval, the principals of public primary schools were contacted via email or phone, and an email containing the MoE and JPN approval letter, the background of the study, procedures of the experiment, information of the experimenter, invitation brochure and questionnaire link that parents would receive, were sent to the schools. Parents who expressed interest were screened via phone for eligibility of their child to participate in the study. The inclusion criteria for the typically developing group and ASC group were the same as the criteria stated in Chapter 2. The inclusion criteria for the TD group comprise no formal diagnosis and not suspected of any developmental, neurological, or psychiatric disorders or a known genetic condition. The inclusion criteria for the ASC group were the child has received a formal diagnosis of ASC by a professional (e.g., medical practitioners, paediatricians), and is absence of brain injury, epilepsy, or a known genetic condition. The questionnaires were available in English, Mandarin and Bahasa Malaysia. The parents could choose their preferred language. The access link to the online questionnaire was sent to parents who met the eligibility criteria and who had provided informed consent. The steps of the recruitment procedures can be found in Appendix 1.

An initial session was carried out with children of parents who completed questionnaires. A timetable with five slots a day was created and sent to the administrator for tracking purposes. Each slot lasted for 30 minutes. During the initial session, children were shown an information sheet with a simple description of the purposes and procedures of the experiment (session 1 and 2) along with pictures related to the content. A sample of the children's information sheet is shown in Appendix 2. Children who agreed to participate in the experiment continued immediately to the first session and the second session on another day. Similar to Chapter 2, the participants could request an interim report regarding the child's ability in Emotion Regulation, Executive Functioning, repetitive behaviours and sensory sensitivity based on the completed questionnaires.

### 3.2.2 Materials and Measures

#### 3.2.2.1 Demographic Information

A sample of demographic questionnaire for this study is provided in Appendix 3.

#### 3.2.2.2 Preference Assessment

Parents were asked to identify five movies or videos that the child liked to watch and neither liked nor disliked strongly and rated their child's preference in toys and activities that were suitable to provide in a classroom setting on a five-point scale ranging from "strongly dislikes" to "strongly likes". The questions used in the indirect preference assessment are provided in Appendix 4.

#### 3.2.2.3 Autistic Traits Measure

Following the poor internal consistency of the AQ-10 (Allison et al., 2012) in the Chapter 2 sample, we used the Autism Quotient -Short (AQ-Short; Hoekstra et al., 2011), another short version of the Autism Quotient (AQ; Baron-Cohen et al., 2001) which contains 28 items selected from the original 50 questions. Each item is answered on a four-point scale, ranging from 1 indicating "definitely agree" to 4 indicating "definitely disagree", with a higher total score indicating more autistic traits. Although we used the AQ-28 (Hoekstra, 2011), which used a 4-point Likert scale, we scored dichotomously in line with the original AQ (Baron-Cohen et al., 2001). The score could hence vary between 0 and 28. Compared to the AQ, the AQ-Short was claimed to have a clearer factor structure, comprising two higher-order factors: the social factor (Social behaviour) and the non-social factor (Numbers or patterns; Hoekstra et al., 2011). The correlation between these two higher-order factors was low (r between .16 and .20), which suggests that they can be considered independent variables. Cronbach's alphas (α) were calculated for the current sample (*n* = 119) and found acceptable internal consistency of .65. This assessment is used to measure the differences of autistic traits between ASC and TD groups.

#### 3.2.2.4 Repetitive Behaviours Measure

The *Repetitive Behaviour Questionnaire-2 (RBQ-2;* Leekam et al., 2007), fully described in Chapter 2, was used to measure parental reports of RRBs as it presented high internal consistency in that study for the Malaysian population (α = .88). Cronbach's alphas (α) were calculated for the current sample (*n* = 120) and found high internal consistency of .86.

#### 3.2.2.5 Executive Functioning Measure

The *Childhood Executive Functioning Inventory (CHEXI;* Thorell & Nyberg, 2008), fully described in Chapter 2, was used to measure Executive Functioning in children in which a high internal consistency (α = .97) was found in the previous chapter. Cronbach's alphas (α) were calculated for the current sample (*n* = 120) and found high internal consistency of .94.

#### 3.2.2.6 Emotion Regulation Measure

The *Emotion Regulation Checklist (ERC;* Shields & Cicchetti, 1997), fully described in Chapter 2, was used to measure children's emotion regulation ability, as a high internal consistency (α = .87) was found in the previous chapter. Cronbach's alphas (α) were calculated for the current sample (*n* = 120) and found acceptable internal consistency of .78.

#### 3.2.2.7 Social Pragmatic Measure

The *Pragmatic Abilities Questionnaire (PAQ;* Jafari et al., 2019) of a Cronbach alpha of .98 was used to measure children's social communication abilities. The measure was fully described in Chapter 2. Cronbach's alphas (α) were calculated for the current sample (*n* = 120) and found high internal consistency of .98.

#### 3.2.2.8 Cognitive Abilities Measure

The *Waschbusch's Parent-Rated General Cognitive Ability Scale* (Waschbusch et al., 2000), fully described in Chapter 2, which presented high internal consistency of .96, was used to measure children's general cognitive ability. Cronbach's alphas (α) were calculated for the current sample ($n$ = 120) and found high internal consistency of .94.

#### 3.2.2.9 Sensory Sensitivity Measure

While most of the questionnaires from Chapter 2 remained in Chapter 3, one questionnaire, the *Sensory Sensitivity Questionnaire (SSQ; Minshew and Hobson, 2008)* was replaced with another questionnaire, the Short Sensory Profile, Second Edition (SSP-2; Dunn, 2014) in measuring the sensory sensitivity of the participants in Chapter 3. This decision was made due to the low internal consistency (Cronbach's alpha; α = .56) in the sample described in Chapter 2.

The SSP-2 provides a brief measurement evaluating sensory processing patterns of children between 3 to 14 years. This questionnaire contains 34 items which are items with the greatest discrimination power from the Child Sensory Profile 2 (W. Dunn, 2014). Each item is answered on a five-point scale, ranging from 1 (*almost never*) to 5 (*almost always*). These scores were also calculated for the Sensory and Behavioural scales, in which the Sensory scale refers to the child's sensory processing, and the Behavioural scale refers to the behavioural responses associated with sensory processing. The scores can also be calculated for the four quadrants of Dunn's Sensory Processing Framework: Seeking/Seeker, Avoiding/Avoider, Sensitivity/Sensor, and Registration/Bystander. The seeking/Seeker quadrant has been defined as "the degree to which a child obtains sensory input," where children with a higher score seek sensory input at a higher rate than others. Avoiding/Avoider refers to "the degree to which a child is bothered by sensory input," children with a higher score move away from sensory input at a higher rate. Sensitivity/Sensor refers to "the degree to which a child detects sensory input," and Registration/Bystander refers to "the degree to which a child did not notice changes in the sensory situation.” Cronbach’s alphas (α) were calculated for the current sample ($n$ = 120) for the Sensory scale and found high internal consistency of .94.

**3.2.2.10 Translation**

All questionnaires were translated into Bahasa Malaysia and Mandarin to target a broader population in Malaysia, a country consisting of three main ethnic communities: Malay (61.8%), Chinese (21.4%) and Indian (6.4%), as of 2015 (The Malaysian Administrative Modernisation and Management Planning Unit, 2022). We emailed the authors of the original questionnaires requesting for Bahasa Malaysia and Mandarin versions of the questionnaire, if available. Questionnaires that were already available in Bahasa Malaysia and Mandarin were not translated, and the information of the authors who conducted the translation into these two languages is summarized in Table 3.2.

For questionnaires that had not been translated from English, they were first translated into Bahasa Malaysia and Mandarin by the author, and then back-translated by native-speaker colleagues of each language. Sixty participants completed the English version of questionnaires, 60 completed the Mandarin version, and no participants completed the Bahasa Malaysia version. The participants were recruited from 4 national schools, 1 international school and 2 private early intervention program (EIP) and special education centres. Therefore, participants are believed to be broadly representative of the population of Malaysia. The first translation and back-translation, together with the original questions, are presented in Appendix 5 (Chinese version) and Appendix 6 (Bahasa Malaysia).

**Table 3.2**

*Table of studies provided translated questionnaires in Bahasa Malaysia (BM) and Mandarin*

| Questionnaire | Authors of BM version | Authors of Mandarin version |
| --- | --- | --- |
| Autism Quotient (AQ) | Chee & de Vries (2021) | Lin et al. (2017) |
| Repetitive Behaviour Questionnaire-2 (RBQ-2) | Not available | Leekam et al. (2007) |
| Childhood Executive Functioning Inventory (CHEXI) | Available on https://chexi.se/downloads. | Thorell et al. (2013) |
| Emotion Regulation Checklist (ERC) | Dzulkarnain et al. (2020) | Guo & Cicchetti (2014) |
| Short Sensory Profile (SSP) | Not available | Not available |
| Pragmatic Abilities Questionnaire (PAQ) | Not available | Not available |
| Waschbusch's Parent-Rated General Cognitive Ability Scale | Not available | Not available |

**3.2.2.11 Go/No-Go Task**

Two neuropsychological tasks were used to measure the three targeted EF constructs (Working Memory, Response Inhibition and Cognitive Flexibility) stated in Chapter 1. The Go/No-Go task was used to measure Response inhibition primarily, and the Dimensional Card Sorting (DCCS) was used to measure Cognitive Flexibility primarily. These two tasks have been found to engage working memory.

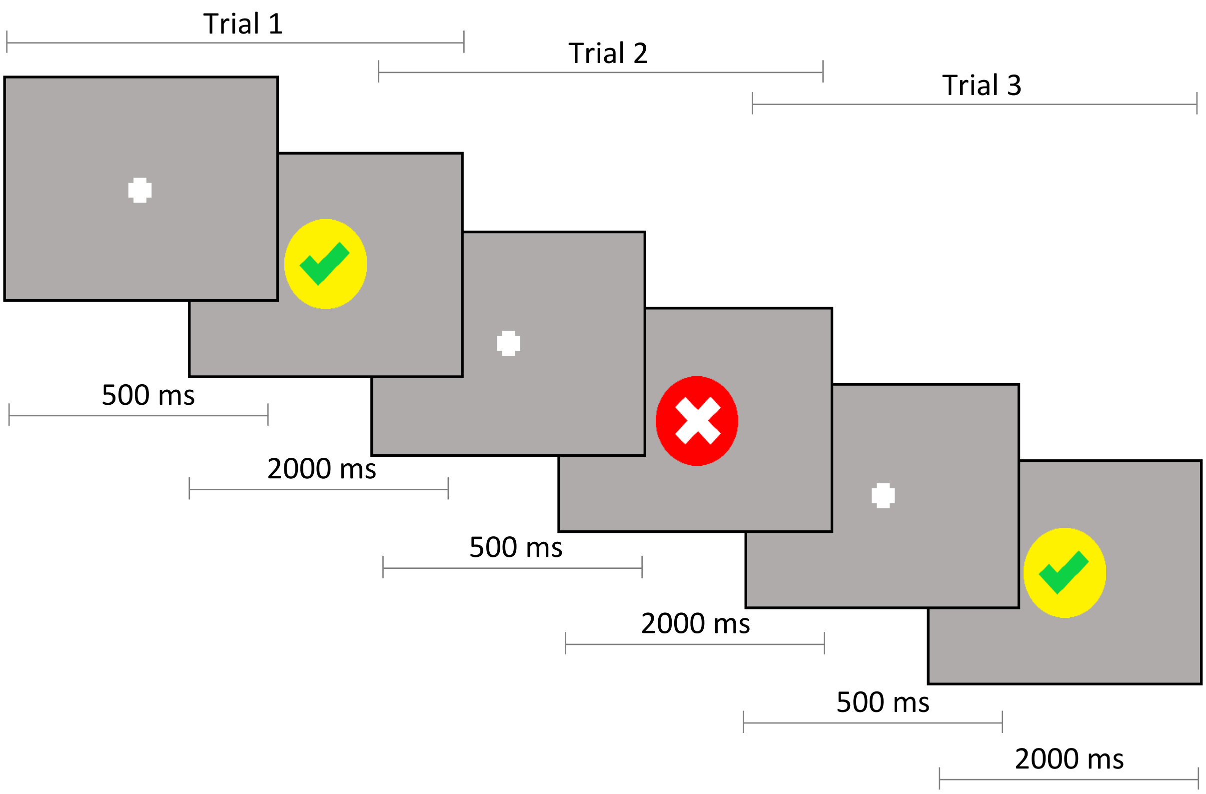
The Go/No-Go task is one of the prominent psychological paradigms commonly used to measure response inhibition (Meule, 2017). The performance on this task has been found to be affected by working memory capacity (WMC) in which individuals with low-WMC perform poorer in this task compared to individuals with high-WMC (Redick et al., 2011). A computerised Go/No-Go task was developed in this study using PsychoPy v.2021.2., an open-source software package for running experiments in Python programming language (Peirce et al., 2019).

The Go/No-Go task consists of Go trials (response) and No-Go trials (inhibition). Participants were told to press the button as fast as possible when the target stimulus (a yellow circle with a green check in the middle) was displayed on the computer screen, and withhold this reaction when they saw the other stimulus (a red circle with a white cross in the middle). Only one yellow button with the size of 9cm x 9cm x 4cm (L x W x H) was placed in front of the participants to minimise distractions during the task. A button with a yellow surface should increase the salience of the cue to press when a yellow circle is presented on the computer screen. These stimuli were displayed against a grey background to make them more salient and to capture the participant's attention (Doebel & Zelazo, 2015).

A white cross (1.5cm x 1.5cm) was used to gain participants' attention before presenting the target stimulus, which was shown for 500 ms (milliseconds) in the middle of the screen, where the stimulus was displayed. The task in this study adapted the Go/No-Go task design from a study targeting children with high functioning autism (HFA) between 8 to 14 years old (Xiao et al., 2012), in which a block of 20 trials was given to the participants in a randomized sequence, of which 25% were No-Go trials. Each trial was displayed for 2000 ms, but the participants could immediately move on to the subsequent trial after pressing the button. Response times and accuracy of performance were recorded using the PsychoPy software. The ratio 3:1 with intertrial 450 ms produces the highest false alarm rate (Young, 2018). A schematic illustration of three sample trials of the Go/No-Go task is provided in Figure 3.1.

**Figure 3.1.**

*Schematic illustration of three sample trials of the Go/No-Go task.*




A ceiling effect was observed in Go/No-Go responses as among the 59 participants with typical development, more than 64% of the participants (n = 38) scored 100% in the Go/No-Go task, resulting in the mean score of 97.46% (*SD* = 4.29, 95% confidence interval [CI] = 96.34, 98.58). In the ASC group, 30% of the participants (n = 15) scored 100%, 17 participants scored 80% and above in the Go/No-Go task, and 17 out of 49 scored below 80%. According to Lim et al. (2015), ceiling effects are considered significant when more than 15% of participants scored the best score (i.e., 100%). Therefore, from the above results, both TD and ASC showed ceiling effects on the Go/No-Go task. Instead of using solely the percentage of correct responses and the response time, the inverse efficiency score (IES; Townsend & Ashby, 1978) was used to analyse the Go/No-Go task performance.

The IES is usually defined as mean reaction time of correct responses divided by proportion of correct responses (Liesefeld & Janczyk, 2019):

$$IES_{i,j} = \frac{RT_{i,j}}{PC_{i,j}}$$

IES refers to inverse efficiency score

RT refers to mean reaction time of correct responses

PC refers to proportion of correct responses

*i* refers to participant *i*

*j* refers to condition *j*

Speed (reaction time) and accuracy (proportions of correct responses) are usually two ideal outcomes of performances in behavioural experiment as they are easy to interpret. Unfortunately, it remains largely unpredictable whether participants will focus more on doing the task quickly or on exhibiting the correct responses. The speed-accuracy trade-off (SAT) suggests an inverse relation between speed and accuracy, in which participants either respond fast but their responses are more prone to error, or sacrifice speed to respond accurately (Heitz, 2014). Therefore, to avoid contradictory findings, reaction time and correct responses are integrated into one measure with the IES.

**3.2.2.12 Dimensional Change Card Sorting (DCCS) Task**

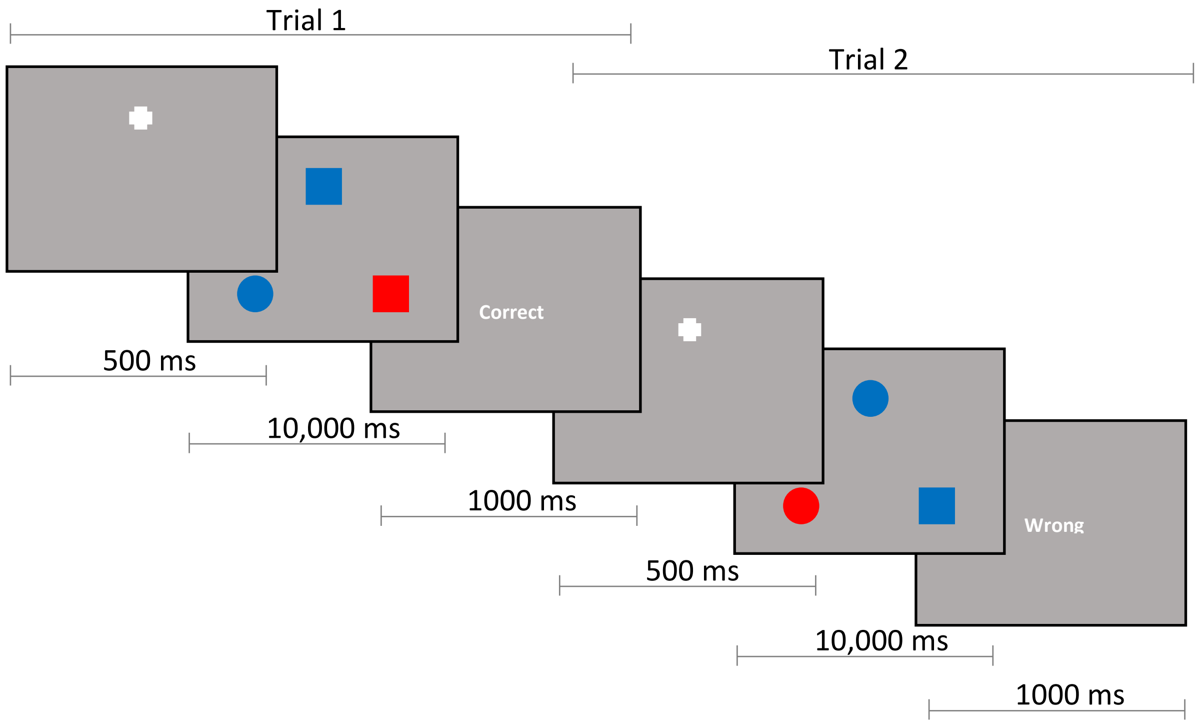
The Dimensional Card Change Sorting (DCCS) test, developed by Frye et al. (1995), has been widely used to measure the Executive Functioning abilities of typically developing and clinical samples. The performance on the DCCS involves cognitive flexibility (to shift the behaviour of matching stimuli according to the alternative dimension from the previous dimension) and other Executive Functioning components such as working memory (to withhold the rules in mind) and response inhibition (to suppress attention to the previous dimension). Similar to the Go/No-Go task, a computerised DCCS task was developed in this study using PsychoPy v.2021.2.3 (Peirce et al., 2019). Participants were asked to match the stimuli by either shape (square or circle) or colour (red or blue) with both verbal and written instructions in the beginning of the session. Simple shapes were used because individuals with ASC have been known to be more biased towards detail (Chung & Son, 2020). The complexity of stimuli affects their visual perception abilities (Bertone et al., 2005), and simple shapes (square and circle) are postulated to minimise perceptual bias. One target stimulus (e.g., a blue square) was presented above two other stimuli (e.g., a blue circle on the left and a red square on the right) against a grey background to make the stimuli more salient and to increase the participant's attention (Doebel & Zelazo, 2015).

In order to reduce the involvement of other cognitive skills such as learning from feedback and identifying changes of rule, the trials of were categorized into test blocks. Participants first completed eight practice trials (or one practice block) prior to five test blocks. The results from the practice block were not included in the data analysis. Each test block consisted of eight trials and used one of the two sorting rules (i.e., match by colour or shape). Blocks 1, 3 and 5 used one rule, and blocks 2 and 4 used the other. The number of pre-switch trials (number of trials in each block) is generally ranged from 1 to 15, with most researchers administering five or six trials in research targeting typically developing children (Doebel & Zelazo, 2015) and ten trials in research targeting children with ASC between 6 to 17 years old (Diamond & Kirkham, 2005; Dichter et al., 2010). According to the active-latent account (Morton & Munakata, 2002), additional pre-switch trials strengthen latent representations of these features, increasing perseveration. The pre-switch trial was pre-determined at eight trials per block, targeting a younger age range (5 to 14 years old) in this study.

Similar to the Go/No-Go task, the task was administered using a laptop computer with a 14” screen, a white cross (1.5cm x 1.5cm) was presented for 500ms in the target stimulus’ position (i.e., the upper part of the screen) to gain participants’ attention before presenting the target and the other two stimuli. The rule (e.g., “Now match the colours”) was presented in written instructions before each block, and participants were told to match the stimuli by pressing either the left button or the right button with each size of 9cm x 9cm x 4cm (L x W x H). They then received performance feedback (i.e., “correct” or “wrong”) following their responses. The stimuli were presented for 1000ms. The feedback was provided and immediately moved on to the subsequent trial if the participants responded within this duration. A schematic illustration of two sample trials of the DCCS task is provided in Figure 3.2.

**Figure 3.2.**

*Schematic illustration of two sample trials of the DCCS task.*




In the DCCS task, we calculated the percentage of correct responses, reaction time and the average transition time, which measures the reaction time difference between the first two trials in each block and the last two trials of the previous block. For the average transition time, Dichter et al. (2010) suggested that the reaction time difference between these blocks involves the performance of set-shifting, which is also known as cognitive flexibility. The mean of these reaction time differences was calculated.

Like Go/No-Go task, the IES of DCCS performance was calculated using the percentage of correct responses and reaction time. In addition, the mean accuracy (percentage of correct responses) of the first two trials of all blocks was calculated, representing the performance of cognitive flexibility. As the DCCS used single-task blocks, the mean accuracy (percentage of correct responses) of the last four trials in all blocks was estimated to represent the performance of working memory. If any of these trials were incorrect, the incorrect value would be removed, and the value from the previous or later correct trial would be taken. For example, if one of the first two trials was incorrect, the next value (third trial) would be taken. If one of the last four trials was incorrect, the previous value (third trial) would be taken. If all the first four or last four data were incorrect, the incorrect data were unusable, and the data set was removed.

**3.2.2.13 Raven's Coloured Progressive Matrices (Raven's CPM)**

Raven's CPM is one of the most frequently used non-verbal tests to measure the ability to think, reason and problem solve abstractly (Kent, 2017) and provides an estimation of fluid intelligence, an aspect of general human intelligence (Cotton et al., 2005). The CPM is created specifically for children between 5 to 11 years of age (Sattler, 1992), and it is an alternative version of Raven's Standard Progressive Matrices (SPM). Advocates of this reasoning task have argued that CMP is the purest measure of fluid intelligence especially for children with intellectual disability (Anderson et al., 1968; Kilburn et al., 1966) and language difficulties (Carver, 1990), which has been viewed as a fair measure of intelligence for children across ethnicities and cultures (Carlson & Jensen, 1981; Lynn et al., 2004). This test consists of three sets (i.e., A, Ab, and B), with 12 items each. The items are sequenced in increasing difficulties within each set. These items are brightly coloured to maintain the participants' attention. A drawing with a missing element is presented on the middle top, with six options presented below the drawing. The participants are asked to select the missing element from the six options. In this study, this task was administered using a touchscreen laptop so the participant could simply select by touching the screen with their finger, with no time limit. The total score of correct trials was computed; one point is given for each correct answer, and the maximum score is 36. A study with a sample of 618 children assessed the internal consistency of the CPM using the Kuder-Richerdson Formula 20 (K-R 20) reported estimates of .89 for participants between 6 to 12 years old (Cotton et al., 2005), ranging from .76 (for 11 years old) to .88 (for 8 and 9 years old), indicating the CPM is a reliable measure.

### 3.2.3 Procedure

The experiment of this study was conducted in three separate sessions: 1) parent-report questionnaires, 2) Task condition, and 3) Neutral and Preferred conditions.

#### 3.2.3.1. Session 1: Questionnaires

The questionnaires consist of seven standardized questionnaires: AQ-Short, RBQ-2, CHEXI, ERC, SSP, PAQ and Waschbusch's Parent-Report Cognitive Abilities. These questionnaires were available in English, Bahasa Malaysia and Mandarin, which could be requested based on the parent's preferences. These questionnaires consist of a total of 152 questions and took approximately 30 minutes to complete. The parent provided consent for their child to participate in the subsequent two experiment sessions and for these sessions to be recorded.

#### 3.2.3.2. Session 2: Task Condition

Children whose parents consented to their participation were led to a classroom with minimal distractions. The child was provided with an information sheet containing a brief introduction of the study and the procedures of session 2 (task condition) and 3 (neutral and preferred conditions). Children were informed their participation was voluntary and they could withdraw anytime during the session, those wanting to continue gave their assent via writing their names to 'sign-up'. None of the participants withdrew themselves from the experiment.

Children were given three tasks during this session: 1) the Go/No-Go task, the Dimensional Change Card Sorting (DCCS) task and Raven's Coloured Progressive Matrices. For the first two tasks, the child was required to press a coloured button (red or yellow) to make an appropriate response. A touchscreen laptop was used for Raven's Coloured Progressive Matrices where the child could select the answer by simply touching the screen with their finger. Participants took more than 10 minutes to complete all the tasks and the duration varied across participants (M = 13.81 minutes, SD = 2.08 minutes). However, we only used 10 minutes to evaluate the percentage of RRBs, to be consistent across conditions, as the other conditions (Preferred and neutral videos) lasted 10 minutes.

During the task condition, there were six children with ASC who left their chair, nine children with ASC got distracted and looked away, and one child with ASC engaged in Self-Injurious Behaviours (i.e., head hitting) during the task. Children who left their chair before completing the tasks were encouraged and redirected back to the chair and complete the tasks. The child who engaged in Self-Injurious Behaviours was given time to regulate their emotions before being redirected to the task. Children who were distracted and not looking at the task were also redirected by pointing at the task and saying, "come, let's finish the task," every 15 seconds. Children requiring rest and comfort breaks were able to take them, and the task was restarted again after the child returned. A summary of actions taken in different situations is presented in Table 3.3.

**Table 3.3.**

*Summary of actions taken (solution) in different situations*

| Situation | Solution |
|---|---|
| Children leave chairs during task | Encourage children to go back to their chairs by saying, “come, let’s finish the task,” pointing at the laptop screen where the task is being displayed and using gestures indicating come here.<br>If the child refuses to do the task, then terminate the task, as their data will be unusable unless all trials are completed. |
| Children get distracted and look at other places or engage in RRBs besides the task for more than 5 seconds | Stand in front of children, point at the task and say, “come, let’s finish the task.”<br>If the child does not provide any responses, lightly touch the child’s hand or gesture the child back to the task every 30 seconds. |
| Children take a break during the task | Discourage children from taking a break until the task has been completed, saying, “We are almost done.” If children need to take a break to the toilet, terminate the task and restart the task again when they are back. Incomplete data will be unusable. |
| Children engage in repetitive behaviours that may lead to harm | Redirect the child from repetitive behaviour to the on-going activity with minimal gesture prompt. |

| | |
|---|---|
| Children request to withdraw from the study | If the child is doing the last task, simply ask, “Are you sure? We are almost done.”<br>If the child is not doing the last task, confirm with the child saying, “Are you sure?”<br>If the child wants to continue, return to the task.<br>If the child confirms withdrawal, terminate the task |

#### 3.2.3.3. Session 3: Neutral and Preferred Conditions

During this session, the child was presented two videos for 10 minutes each. During the Neutral condition, the child was shown a video that had been suggested by the parent, which the child neither liked nor disliked. During the Preferred condition, the child chose a video to watch. Children who were unable to choose were shown one suggested as positive by their parents. Five children with ASD who did not want to watch a video were given preferred toys and activities as proposed by the parents. These two conditions were counterbalanced and pseudo-randomly assigned to all children: 55 children had the neutral condition first, and the other 56 children had the preferred condition first. A 10-minute timer was started to indicate the onset of each condition and when the timer rang, the video was stopped, or the toys were removed. Both session 2 and 3 were recorded with a camera and after explaining the instructions, the experimenter moved at least one meter away from the child during the movie/activity time.

#### 3.2.3.4. Set-Up

There were two tables, two chairs, a tripod holding a camera, a laptop and two buttons used in session 2 and 3. An overview of onsite set-up is provided in Figure 3.3.

**Figure 3.3.**

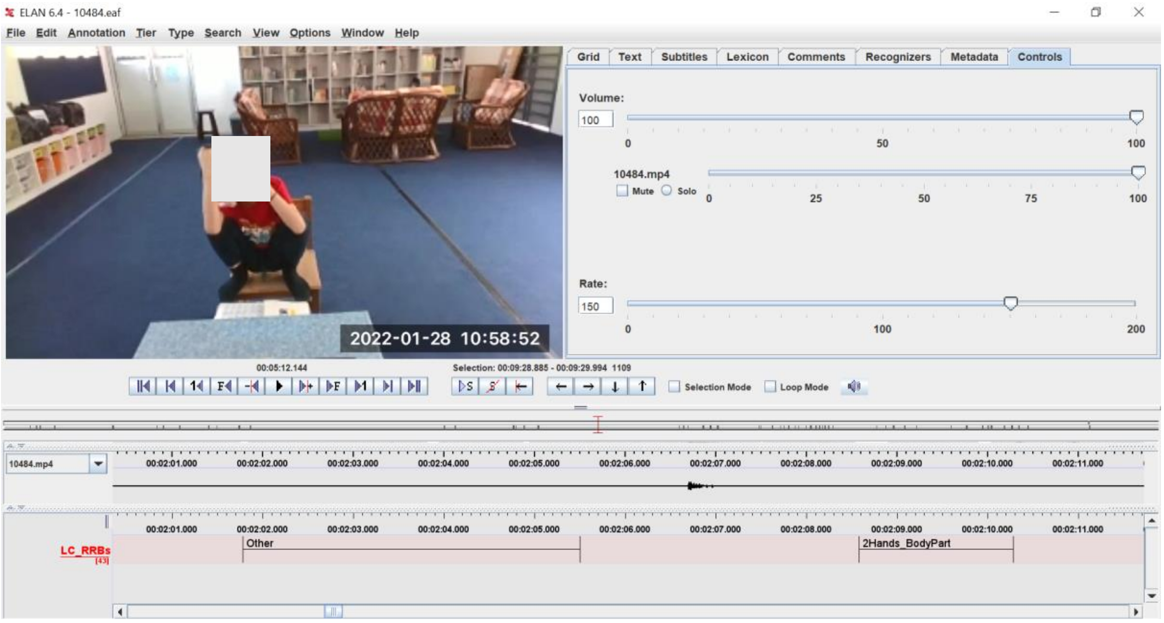
*On-site session set-up*

### 3.2.4 Video Analysis and Behavioural Coding

The participants' RRBs were coded with ELAN (Version 6.4) [Computer software, 2022], a computer software that provides an annotation function for video recordings (Lausberg & Sloetjes, 2009). Figure 3.4 shows a screenshot of an annotated video in the ELAN environment. To our knowledge, this study was the first to investigate repetitive behaviours by recording the behaviours of children between 5 to 13 years old across three contexts: Task, Preferred and Neutral conditions.

**Figure 3.4**

*Behavioural coding in the ELAN environment*




These behaviours were coded based on a list of operational definitions of RRBs described in Table 3.5. The operational definitions were developed referring to the Repetitive Movement and Restricted Interest Scales (RMRIS; Stronach & Wetherby, 2014) that categorized movements involving objects and body parts. In this thesis, we focused only on movements that involved arms and hands. Three types of situations were created, with: 1) "Only" indicating movements that involved either arm, arms, or hands only without touching the surface of any non-living things nor body parts, 2) "Object" indicating movements that involved either arm, arms, or hands touching the surface of non-living things, and finally, 3) "BodyPart" indicating movements that involved either arm, arms, or hands touching the surface of any body part. A controlled vocabulary was created and added to the list of behavioural codes in the behavioural coding software, ELAN (Version 6.4), and the template was standardized and used for all video analyses.

**Table 3.5.**

*Operational definitions of RRBs*

| Code | Definition |
|---|---|
| Arm_Only | Any non-contextual behaviour involving one arm moving in any direction repeatedly for 2 or more cycles without touching any surface of non-living thing or body part, with no more than 1 second between cycles. Example, engage in hand-flapping during task. Non-example, raising hand upon instructions. |
| 2Arms_Only | Any non-contextual behaviour involving both arms moving in any direction repeatedly for 2 or more cycles without touching any surface of non-living thing or body part, with no more than 1 second between cycles. Example, engage in hand-flapping during task. Non-example, raising both hands upon instructions. |
| Arm_Object | Any non-contextual behaviour involving one arm touching the surface of any non-living thing in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles. Example, tapping on table during task. Non-example, pressing button to perform the task. |
| 2Arms_Object | Any non-contextual behaviour involving both arms touching the surface of any non-living thing in any direction repeatedly for 2 or more cycles, with no more |

| | |
|---|---|
| | than 1 second between cycles. Example, tapping on table during task. Non-example, pressing on buttons during task. |
| Arm_BodyPart | Any non-contextual behaviour involving one arm touching the surface of any body part in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles. Example, stretching upon completion of task. Non-example, scratching head once due to itch. |
| 2Arms_BodyPart | Any non-contextual behaviour involving both arms touching the surface of any body part in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles. Example, clapping hands during task. Non-example, clapping hands upon instructions. |
| Hands_Fingers_Only | Any non-contextual behaviour involving one or both hands, or any fingers moving in any direction repeatedly for 2 or more cycles without touching any surface of non-living thing or body part, with no more than 1 second between cycles and without apparent displacement. Example, flickering fingers during task. Non-example, waving to get experimenter's attention. |
| Hands_Fingers_Object | Any non-contextual behaviour involving one or both hands, or any fingers touching surface of any non-living thing in any direction repeatedly for 2 or more cycles, with no |

| | |
|---|---|
| | more than 1 second between cycles. Example, tapping fingers on table during task. Non-example, spinning fidgety spinner. |
| Hands_Fingers_BodyPart | Any non-contextual behaviour involving one or both hands, or any fingers touching the surface of any body part in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles. Example, scratching the same part of head repeatedly during task. Non-example, scratching hand during task. |
| Other | Any non-contextual behaviour involving other body parts besides arms, hands and fingers, with or without touching the surface of any non-living things or any body part in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles. Example, shaking leg repeatedly during task. Non-example, standing up and leaving the room after finishing the task. |

### 3.2.5 Data Analysis

The statistical analyses were conducted using IBM SPSS Statistics for Windows, Version 25.0. Missing data were replaced with the series mean method if the data set within a questionnaire has less than 25% missing data. The mean of the non-missing values of the participant in the target instrument was calculated and then replaced the missing values separately and independently from other participants within the same instrument. Listwise deletion was used if there were more than 40% missing data (a rule of thumb suggested in

(Jakobsen et al., 2017) in the participant's questionnaire. The percentage of missing data for all measures is summarised in Appendix 7.

RRBs durations were hypothesized to be longer in conditions involving emotional arousals such as Task and Preferred conditions relative to Neutral conditions, especially for children with poorer Emotion Regulation. A one-way ANOVA was conducted to compare RRBs duration across these conditions. Similar to the findings from Chapter 2, we hypothesized that Emotion Regulation serve as a mediator in the relationship between Executive Functioning and RRBs in the total sample and ASD group. To test this hypothesis, first we examined the normality of all variables with Shapiro-Wilk tests which indicated normal distribution for the CHEXI and ERC but not for RBQ-2, Waschbusch's Parent Report Cognitive Scale, SSP, PAQ, Go/No-Go IES, and DCCS IES and DCCS average transition time. For the analyses, a two-step procedure: 1) transformation to uniformity and 2) transformation to normality (Templeton, 2011) were performed on the latter variables due to violations in normality. Then, the response differences across TD and ASC groups were examined with an Independent two-tailed t-test and the mediating roles of Emotion Regulation was tested with mediation analysis, Hayes' PROCESS model 4. The bootstrapping method was used to analyse the standard errors of the data set (Hayes & Rockwood, 2020).

#### 3.2.5.1 Interobserver Agreement

Two coders were trained to code objectively based on the given operational definitions with 20 training videos that are not from the current study. A mutual discussion was conducted after every training video and interobserver agreement was also calculated. Interobserver agreement for the occurrences and non-occurrences of RRBs measured by duration was calculated using Cohen's kappa (Cohen's κ). Cohen's κ measures the reliability of categorical scales by assessing the agreement between observers and has a value ranging from 0 to 1. Values between .81 and 1.00 indicate almost perfect agreement, .61 to .80 as substantial, .41 to .60 as moderate, .21 to .40 as fair, .01 to .20 as none to slight, and values less than or equal to 0 as no agreement (Cohen, 1960). In the final 15 training videos, all items reached interobserver reliability for at least .80 and coders proceeded to coding the videos of the current study.

During the actual video coding of the current study, all videos from the TD and ASC groups were mixed and randomized to ensure coders were blind to the participants' diagnoses while coding. Both coders coded the behaviours independently, and interobserver agreement was calculated on 25% of the data using 29 randomly selected participants, 11 from the ASC and 18 from the TD groups, respectively. These videos were selected in advance and coders were blind to which participants were included for reliability. A two-by-two contingency table was constructed to calculate the agreement between two coders relative to the duration of any RRBs versus no RRBs. A kappa of .93 was obtained, indicating good agreement overall for the duration of RRBs.

#### 3.2.5.2 Composite Variable

The z score of Go/No-Go inverse efficiency score (GNG_IES), DCCS inverse efficiency score (DCCS_IES) were averaged to create a composite variable of neuropsychological tests of Executive Functioning (C_EF).

#### 3.2.5.3 Standardization

Annotation statistics were extracted from the ELAN environment, which includes the annotation duration percentage and the total annotation duration. The annotation duration percentage refers to the ratio between the total annotation duration of RRBs, and the total recording duration expressed as a percentage. The total annotation duration refers to the total duration of the annotated RRBs. One of the most widely used python data analysis libraries that expedite the data analysis process, Python’s Panda library functions (The pandas development team, 2020) was used to compile the huge amount of extracted annotation statistics to reduce human error possibly caused by manual data entry.

As mentioned above, this study has three conditions: Task, Preferred and Neutral. The Preferred and Neutral conditions were 10 minutes each. However, the duration of the Task condition varied depending on the duration participants took to complete three tasks (i.e., Go/No-Go, DCCS and Raven’s CPM). The duration of all RRBs (in percentage) were converted into z-scores using the mean across all conditions, as presented in the following formula:

$$Z = \frac{x - \bar{x}}{\sigma}$$

Z refers to the standard score

x refers to observed value

$\bar{x}$ refers to the mean of RRBs (in percentage) across all conditions

σ refers to the standard deviation of the sample across all conditions

## 3.3 Results

### 3.3.1. Group Analysis

As mentioned in the Method section, 19 children from the TD group were attending remedial classes for additional academic support. These children were grouped as TD_R and analysed separately from the TD group to examine the risk of bias. A t-test was used to examine the differences between TD and TD_R groups. The results are presented in Table 3.6.

**Table 3.6.**

*Differences between TD and TD_R in autistic traits, Executive Functioning, Emotion Regulation and RRBs.*

| Measure | *M (SD)* | | *p value* |
|---|---|---|---|
| | TD (*n* = 41) | TD_R (n = 19) | |
| Short AQ | 11.78 (3.68) | 11.99 (4.55) | .98 |
| CHEXI Total Score | 66.39 (15.14) | 74.89 (16.60) | .14 |
| Go/No-Go IES | .46 (.59) | .75 (.50) | .24 |
| DCCS IES | -13.09 (24.03) | 4.34 (17.65) | .07 |
| Composite_EF | -.95 (1.81) | -.29 (1.35) | .39 |
| ERC Total Score | 68.82 (6.85) | 67.36 (8.49) | .78 |
| RBQ-2 Total Score | 26.83 (6.20) | 25.87 (6.38) | .84 |
| RRBs Duration | 4.53 (7.53) | 4.69 (6.04) | 1.00 |

There was no significant difference in the mean score between TD and TD_R groups in the four main domains: autistic traits, Executive Functioning, Emotion Regulation and RRBs. Based on the above results, it is believed that it is appropriate to analyse children from the TD and TD_R children together in one group. The mean comparison of parent-report measures between TD, TD_R and ASC groups can be found in Appendix 8.

### 3.3.2. Descriptive Statistics

A series of independent samples t-tests and Mann-Whitney U tests (for measures that violated normality) were performed to compare ASC and TD groups (see Table 3.8).

As mentioned previously, we expected significant differences in autistic traits, RRBs, Executive Functioning and Emotion Regulation but no difference in IQ. For autistic traits, parents of ASC children reported significantly higher mean scores than parents with TD children. For the RRBs domain, parents indicated that children with ASC had significantly more RRBs in the RBQ-2 and significantly longer duration of RRBs were observed in children with ASC in relative to TD children. Children with ASC were also reportedly having significantly higher mean score in the CHEXI, which indicated weaker Executive Functioning than TD children. This pattern of results was similar in the neuropsychological measures. Children with ASC had a significantly higher IES in Go/No-Go task as well as DCCS compared to TD children. The higher the indicator refers to the lower the performance accuracy over time. TD children also had higher mean score in the ERC, which indicated stronger Emotion Regulation than children with ASC. For sensory sensitivity, children with ASC scored significant higher in behavioural section but no difference in sensory section in SSP-2 between children with ASC and TD children. The score of behavioural section reflects the child's behavioural responses associated with sensory processing, whereas sensory section reflects the child's sensory processing abilities. The PAQ scores were also significantly different between ASC and TD groups. There was no difference between children with ASC and TD children in IQ based on the parent-report *Waschbusch's Parent-Rated General Cognitive Ability Scale* and their performances in Raven's CPM.

**Table 3.8.**

*Descriptive statistics of all parent-reports, and observational and performance-based measures across TD and ASC groups*

| Domains | Subdomains and Total | *M (SD)* | | *Independent t-test or Mann-Whitney U tests* | | |
|---|---|---|---|---|---|---|
| | | TD (*n* = 60) | ASC (*n* = 54) | *t* value | *df* | Sig (two-tailed) |
| Age | | 9.77 (22.93) | 10.05 (22.39) | 1512.50 | | .542 |
| Autistic Trait | | 11.85 (3.94) | 14.89 (3.85) | -4.16 | 112 | <.001 |
| Restricted and Repetitive Behaviours (RRBs) | Repetitive Sensory/Motor | 12.08 (3.32) | 14.17 (3.33) | -3.34 | 112 | .001 |
| | Insistence on Sameness | 11.03 (2.55) | 13.63 (3.35) | -4.69 | 112 | <.001 |
| | RBQ2 Total Score | 26.23 (5.98) | 31.58 (6.08) | -4.53 | 112 | < .001 |
| Duration of RRBs (in percentage) | Total Occurrences | 4.58 (7.03) | 9.14 (5.55) | .146 | 106 | .006 |
| Executive Functioning (EF) | CHEXI Working Memory | 34.64 (9.07) | 42.63 (9.88) | -4.50 | 112 | < .001 |
| | CHEXI Response Inhibition | 34.43 (8.06) | 38.07 (7.68) | -2.46 | 112 | .015 |
| | CHEXI Total Score | 69.08 (15.98) | 80.70 (16.51) | -3.82 | 112 | < .001 |
| | Go/No-Go IES | .55 (.57) | 1.04 (.73) | -3.87 | 105 | < .001 |
| | DCCS Average Transition Time | .92 (3.91) | .61 (1.68) | .000 | 100 | 1.00 |

| | | | | | | |
|---|---|---|---|---|---|---|
| | DCCS IES | -7.68 (23.54) | 23.33 (32.70) | -5.71 | 106 | < .001 |
| Emotion Regulation (ER) | Lability/Negativity | 23.89 (3.13) | 21.52 (3.81) | 3.64 | 112 | < .001 |
| | Emotion Regulation | 44.42 (5.98) | 42.03 (6.15) | 2.10 | 112 | .038 |
| | ERC Total Score | 68.36 (7.37) | 63.56 (8.44) | 3.24 | 112 | .002 |
| Sensory Behaviours | Sensory Section | 26.41 (8.11) | 29.78 (8.82) | -1.98 | 111 | .050 |
| | Behavioural Section | 38.64 (12.57) | 47.28 (14.71) | -3.36 | 111 | .001 |
| Intelligence Quotient (IQ) | Waschbusch's Scale | 14.27 (11.46) | 27.32 (11.61) | .010 | 110 | .992 |
| | Raven's CPM | 72.13 (18.58) | 49.55 (29.47) | -.067 | 106 | .946 |
| Social Pragmatic | PAQ Total Score | 156.53 (42.96) | 117.41 (47.69) | 4.504 | 111 | <.001 |

### 3.3.3. Duration of RRBs across Conditions

One of the main objectives in this Chapter is to explore the duration of RRBs (in percentage) across three different conditions (i.e., Task, Preferred, Neutral). First, the differences of the percentage of RRBs duration between TD and ASC groups across these three conditions were as presented in Table 3.9.

**Table 3.9.**

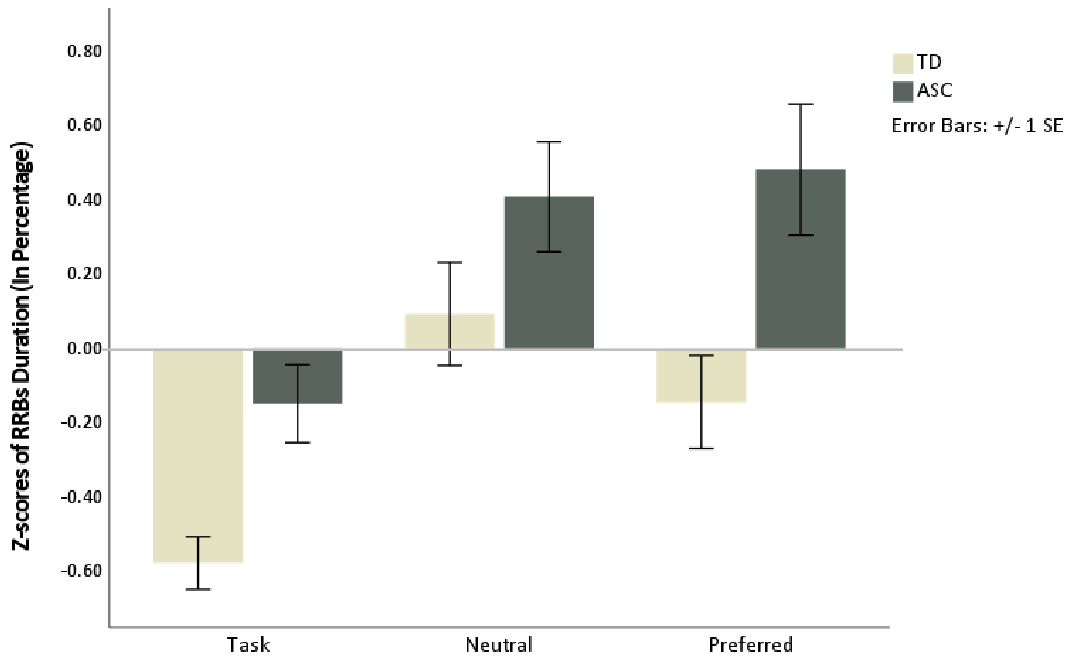
*Descriptive statistics of duration of RRBs (in percentage).*

| Conditions | *M (SD)* | | |
|---|---|---|---|
| | TD (*n* = 57) | ASC (*n* = 50) | Total (*n* = 107) |
| Task | 1.35 (4.63) | 5.14 (6.35) | 3.10 (5.79) |
| Neutral | 7.22 (9.18) | 9.98 (9.07) | 8.50 (9.19) |
| Preferred | 5.15 (8.26) | 10.62 (10.79) | 7.68 (9.85) |

With the use of duration of RRBs (in percentage), TD children engaged in repetitive movements most frequently during Neutral condition, followed by Preferred and Task conditions, whereas children with ASC engaged in repetitive movements most frequently during Preferred, Neutral and finally Task conditions. As mentioned previously, the durations of Preferred and Neutral conditions were pre-set as 10 minutes each, but the duration of Task condition varied based on how quickly the participants completed the tasks. The duration of task condition was significantly related to the percentage of RRBs duration, $r = .27$, $p = .006$ and thus, the varied duration might result in confounding variables. Therefore, as presented previously in *3.2.5.3 Standardization*, all data across three conditions were converted into z-scores using the mean of all conditions. Figure 3.5 showed the results comparing the z-scores of the Task, Neutral and Preferred conditions between TD and ASC groups.

**Figure 3.5.**

*Differences of z-scores between TD and ASC across Task, Neutral and Preferred conditions.*




Note. *$p$ < .05, **$p$ < .01, ***$p$ < .001

A repeated-measures ANOVA was performed to compare the effect of conditions on the z-scores of RRBs duration across TD and ASC. There was a significant main effect of condition on participants' RRBs duration, $F(2, 90) = 28.01$, $p < .001$, $\eta 2 = .384$. There was a significant main effect of group on participants' RRBs duration, $F(1, 45) = 10.02$, $p = .002$, $\eta 2 = .18$. The interaction effect between conditions and group was not significant, $F(2, 90) = 37.92$, $p = .45$, $\eta 2 = .02$.

In the total sample, there was a significant difference across conditions, $F(2, 319) = 12.69$, $p < .001$. There was also a significant difference across conditions in the TD group, $F(2, 171) = 8.82$, $p < .001$, and in the ASC group, $F(2, 147) = 5.64$, $p = .004$. TD children displayed more RRBs during the Neutral condition, whereas children with ASC displayed more RRBs during the Preferred conditions. The differences of RRBs duration between TD and ASC groups across three conditions are shown in Figure 3.6.

### 3.3.4. The mediating effect of Emotion Regulation

Hayes' PROCESS mediation analysis was conducted twice to examine the mediating effect of Emotion Regulation in the relationship between Executive Functioning and RRBs, which is our second objective in this Chapter. The first mediation analysis replicated Chapter 2 and included only the results from the parent-report rating scales, and the second mediation analysis provided an extension from Chapter 2 and included neuropsychological task performances for Executive Functioning domain and observational measures for RRBs domain.

The results from the first mediation analysis are presented in Table 3.10. Mediation diagrams illustrating the relationship between the study variables when all variables measured by parent-report questionnaires are presented in Figure 3.6.

**Table 3.10.**

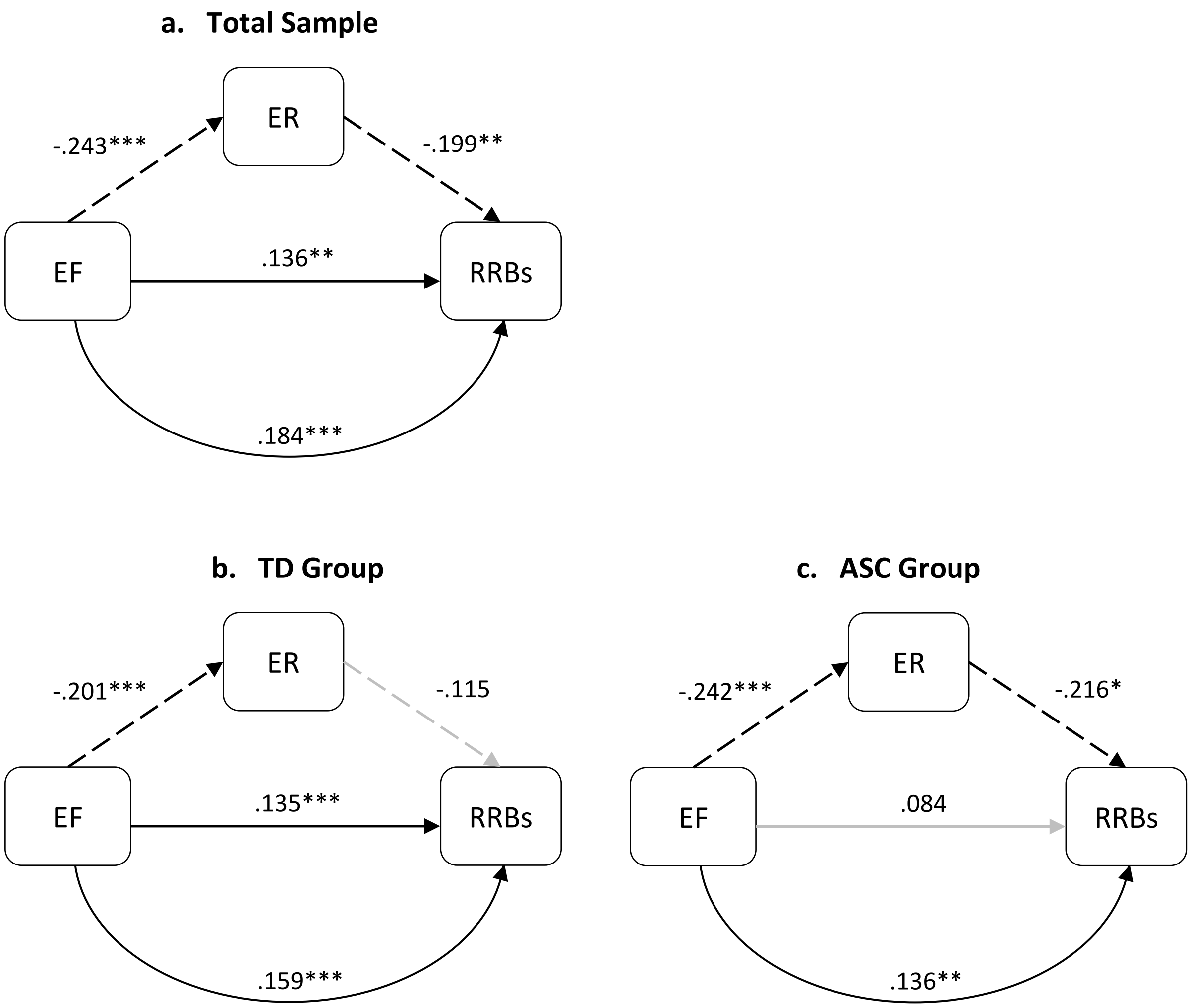
*Mediating effect of Emotion Regulation (measured by ERC) in the relationship between Executive Functioning deficits (measured by CHEXI) and RRBs (measured by RBQ-2).*

| | Total (*n* = 114) | TD (*n* = 60) | ASC (*n* = 54) |
|---|---|---|---|
| a | -.243* | -.201* | -.242* |
| b | -.199* | -.115 | -.216* |
| Direct effect, c’ | .135* | .136* | .084 |
| Total effect, c | .184* | .159* | .136* |
| Indirect effect, a*b | .049* | .023 | .052* |
| 95% CI | [.017, .085] | [-.024, .070] | [.011, .112] |

*Note:* The significance of the effects was assessed through the 95% bootstrap confidence interval (CI). The asterisk*, represents a significant result, i.e., the 95% bootstrap CI does not contain zero. The total effect, c, is the sum of direct effect, c’, and indirect effect, a*b.

**Figure 3.6.**

*Mediation diagrams illustrating the relationship between the study variables (i.e., Executive Functioning (EF), Emotion Regulation (ER) and RRBs) with parent-reports.*




*Note: solid black line represents positive significant relationship, dotted black line represents negative significant relationship, grey line represents non-significant relationship*

*Note.* * $p < .05$, ** $p < .01$, *** $p < .001$

Similar to the findings from Chapter 2, a significant indirect effect from Executive Functioning to Emotion Regulation and finally to RRBs, was found in the total sample and the ASC sample. The total effect, which reflects the sum of direct and indirect effects, was also significant in both groups. These findings indicated that Emotion Regulation serve as mediator in the total sample and the ASC sample.

The findings for TD groups were also similar to the findings from Chapter 2 except the direct effect from Executive Functioning to RRBs in which it was significant in this study. A significant effect of Executive Functioning on Emotion Regulation, and a significant total effect from Executive Functioning to RRBs were found. However, the indirect effect from Emotion Regulation to RRBs was not significant. These findings indicate the relationship between Executive Functioning and RRBs might be mediated by other factors besides Emotion Regulation.

In the second mediation analysis, the relationship between the composite variable of Executive Functioning neuropsychological tests and RRB duration (in percentage) during the Neutral condition was evaluated. We used observation data from the Neutral condition only, instead of Task or Preferred condition, to control for potential valence confounds when examining relationships between Emotion Regulation and behaviours believed to occur as a result of emotion. The results of this mediation analysis are presented in Table 3.11 and Figure 3.8.

**Table 3.11.**

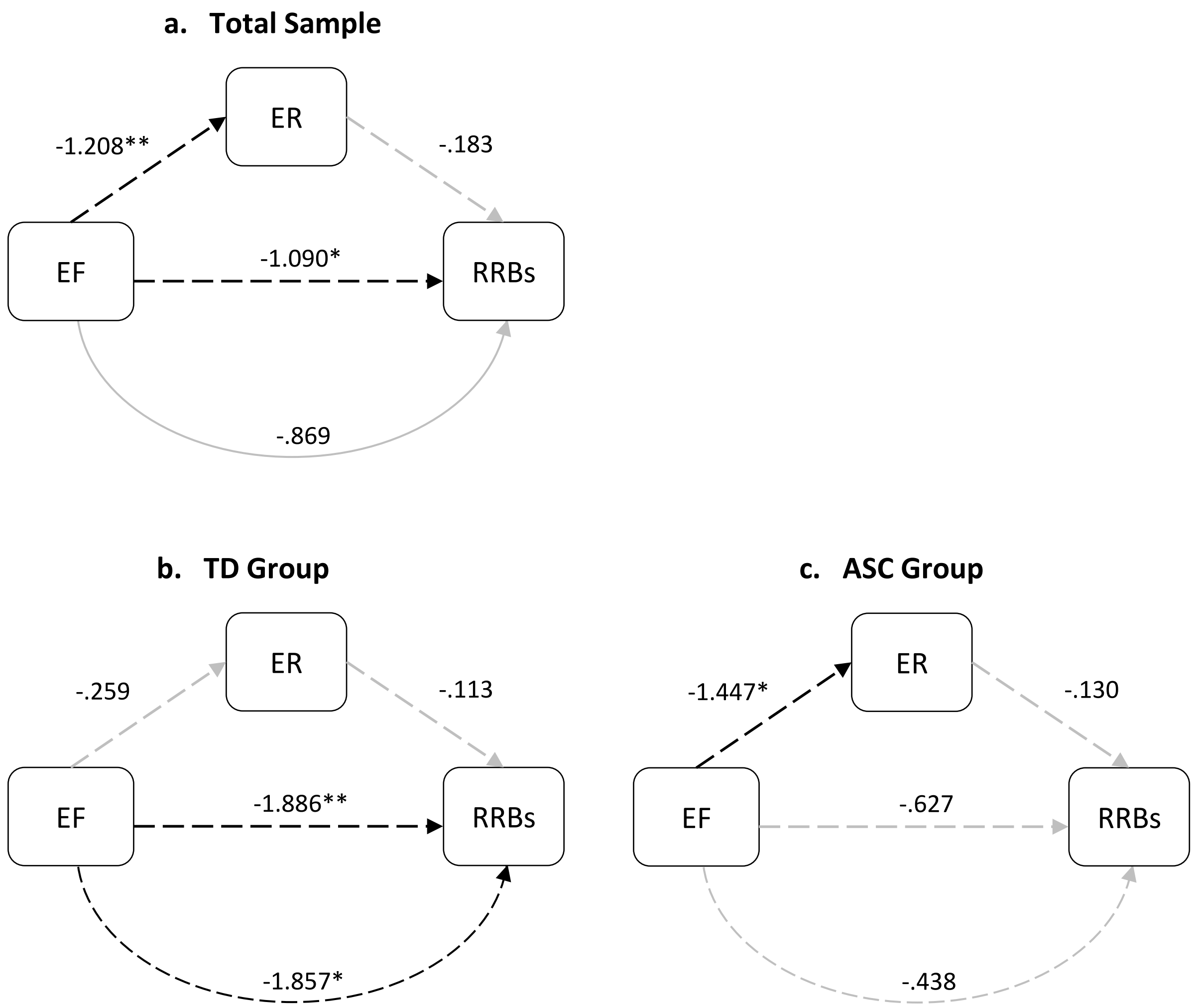
*Mediating effect of Emotion Regulation (measured by ERC) in the relationship between Executive Functioning (measured by Executive Functioning neuropsychological tests) and RRBs (measured using direct observation during Neutral condition).*

| | Total (*n* = 97) | TD (*n* = 55) | ASC (*n* = 42) |
|---|---|---|---|
| a | -1.208** | -.259 | -1.447* |
| b | -.183 | -.113 | -.130 |
| Direct effect, c' | -1.090* | -1.886** | -.627 |
| Total effect, c | -.869 | -1.857* | -.438 |
| Indirect effect, a*b | .222* | .029 | .188 |
| 95% CI | [.003, .609] | [-.122, .433] | [-.199, .831] |

*Note:* The significance of the effects was assessed through the 95% bootstrap confidence interval (CI). The asterisk*, represents a significant result, i.e., the 95% bootstrap CI does not contain zero. The total effect, c, is the sum of direct effect, c', and indirect effect, a*b.

**Figure 3.7.**

*A mediation diagram illustrating the relationship between the study variables when Executive Functioning was measured by two neuropsychological tasks (i.e., Go/No-Go and DCCS) and RRBs were assessed using direct observation during Neutral condition.*




*Note: solid black line represents positive significant relationship, dotted black line represents negative significant relationship, grey line represents non-significant relationship*

*Note.* * $p < .05$, ** $p < .01$, *** $p < .001$

In the total sample, there was a significant effect of Executive Functioning on Emotion Regulation and a significant direct effect of Executive Functioning on RRBs during Neutral condition. In the ASC group, there was only a significant effect from Executive Functioning to Emotion Regulation. In the TD group, there was a significant direct effect and total effect from Executive Functioning to RRBs. However, there was no significant effect of Executive Functioning on Emotion Regulation in the TD group. This was the first occasion across all mediation analyses in Chapter 2 and Chapter 3 that there was no significant effect from Executive Functioning to Emotion Regulation.

In the next section, we will first explore the relationship between Executive Functioning and Emotion Regulation across TD and ASC children. Then, we will explore the pattern of findings between parent-report and neuropsychological or observational-based measures. These analyses were conducted to further examine the individual relationships between Executive Functioning and Emotion Regulation to understand how they affect the mediation analyses, as well as how different measures affect the patterns of findings.

### 3.3.5. Executive Functioning and Emotion Regulation

A Pearson's correlation was conducted to explore the relationship between Executive Functioning and Emotion Regulation (see Table 3.12).

**Table 3.12.**

Pearson relationship between the composite variable of Executive Functioning and ERC.

| | Composite Variable of Executive Functioning | | |
|---|---|---|---|
| | Total Sample | TD | ASC |
| ERC Total Score | -.45*** | -.25 | -.51** |
| Whilst controlling for IQ | | | |
| ERC Total Score | -.37*** | -.17 | .33* |

*Note.* * $p < .05$, ** $p < .01$, *** $p < .001$

A Pearson's correlation coefficient showed that the total score of ERC was significantly related to the composite variable of Executive Functioning, $r(99) = -.45$, $p < .001$, in the total sample. In the ASC group, the total score of ERC was also related to the composite variable of Executive Functioning, $r(43) = -.51$, $p = .001$. In TD group, the total score of ERC was not related to the composite variable of Executive Functioning, $r(56) = -.25$, $p = .07$. This finding explains the second mediation analysis for TD group in which no effect was found from Executive Functioning and Emotion Regulation.

As both Waschbusch's parent-rated general cognitive ability scale and Raven's CPM were related with Executive Functioning measures, a partial correlation was conducted to determine the relationship between the total score of ERC and the composite variable of Executive Functioning, whilst controlling for the total score of Waschbusch's Cognitive Ability Scale and Raven's CPM. In the total sample, there was a statistically significant relationship between the total score of ERC and the composite variable of Executive Functioning, $r(99) = -.37$, $p < .001$. In the TD group, there was no significant relationship between the total score of ERC and the composite variable of Executive Functioning. In the

ASC group, there was a significant relationship between the total score of ERC and the composite variable of Executive Functioning, $r(43) = .33$, $p = .04$. The relationship between Executive Functioning and Emotion Regulation remained significant in the total sample and ASC group whilst both IQ parent-rating scales and neuropsychological tasks were controlled.

### 3.3.6. Parent-Report and Observational-based Measures: RRBs

A Pearson correlation coefficient was computed to assess the linear relationship between observational and rating scale measure of RRBs. There was no significant relationship between the total score of RBQ-2 and the total duration of RRBs in the total, TD and ASC samples. Interestingly, when the relationships were investigated separately in each condition in the total sample, there was a positive significant relationship between the total score of RBQ-2 and RRBs duration during the preferred condition ($r = .20$, $p = .047$) but not during the task ($r = .18$, $p = .07$) and neutral ($r = .10$, $p = .31$) conditions. In the TD and ASC samples, there was no significant correlation between the total score of RBQ-2 and the RRBs duration (in percentage) across all three conditions. The relationships between the parent-report RBQ-2 and observed RRBs duration (in percentage) are summarized in Table 3.13.

**Table 3.13.**

*Pearson correlation between the RBQ-2 (total score and subscales score) and the total duration of RRBs (in percentage), and their duration in each condition.*

| | | RRBs Duration (in percentage) | | | |
|---|---|---|---|---|---|
| | | Total | Task | Neutral | Preferred |
| RBQ-2 (Total Score) | Total Score | .12 | .13 | .10 | .20* |
| RRBs Duration (in percentage) | Total | 1 | . 55*** | .86*** | .90*** |
| | Task | | 1 | .27** | .44*** |
| | Neutral | | | 1 | .61*** |
| | Preferred | | | | 1 |

*Note.* * $p < .05$, ** $p < .01$, *** $p < .001$

### 3.3.7. Parent-Report and Neuropsychological Measures: Executive Functioning and Intelligence Quotient (IQ).

A Pearson correlation coefficient was computed to examine whether the relationship between the parent-rated Executive Functioning rating scale, CHEXI, and the composite variable of neuropsychological tests of Executive Functioning (C_EF). This relationship was not significant (r = .12, $p = .22$), suggesting that the rating measures of Executive Functioning might assess different Executive Functioning construct in relative to the neuropsychological measures of Executive Functioning. A summary of Pearson correlation coefficient between the total and Working Memory (CHEXI_WM) and Response Inhibition (CHEXI_RI) subscales score of CHEXI, the average transition time of DCCS (DCCS_TT), the inverse efficiency score

(IES) of the Go/No-Go (GNG_IES) and DCCS (DCCS_IES) tests, together with the composite variable of EF (C_EF), were presented in Table 3.14 for reference.

A Pearson correlation coefficient indicated that the parent-report measure of IQ was strongly related with parent-report measure of Executive Functioning (r = .65, $p < .001$) but not neuropsychological measure of Executive Functioning. The neuropsychological measure of IQ was strongly related with the composite variable of neuropsychological measures of Executive Functioning (r = -.37, $p < .001$). The results are presented in Table 3.15.

**Table 3.14.**

*Pearson correlation between the total and Working Memory (CHEXI_WM) and Response Inhibition (CHEXI_RI) subscales score of CHEXI, the average transition time of DCCS (DCCS_TT), the inverse efficiency score (IES) of the Go/No-Go (GNG_IES) and DCCS (DCCS_IES) tests, together with the composite variable of EF (C_EF).*

| | CHEXI Total | CHEXI_WM | CHEXI_RI | GNG_IES | DCCS_IES | DCCS_TT | C_EF |
|---|---|---|---|---|---|---|---|
| CHEXI Total Score | - | .95*** | .92*** | .168 | .310** | -.17 | .12 |
| CHEXI Working Memory (WM) | .95*** | - | .76*** | .18 | .35*** | -.16 | .16 |
| CHEXI Response Inhibition (RI) | .92*** | .76*** | - | .13 | .21* | -.15 | .06 |
| GNG_IES | .17 | .18 | .13 | - | .66*** | -.02 | .79*** |
| DCCS_IES | .31** | .35*** | .21* | .66*** | - | -.01 | .79*** |
| DCCS_TT | -.17 | -.16 | -.15 | -.02 | -.01 | - | .48*** |
| Composite Variable of EF (C_EF) | .12 | .16 | .06 | .79*** | .79*** | .48*** | 1 |

*Note.* *p < .05, **p < .01, ***p < .001

**Table 3.15.**

*Correlation between the total score of Waschbusch's Parent-Rated General Cognitive Ability Scale and Raven's Coloured Progressive Matrices (Raven's CPM) and executive functioning measures.*

| | Waschbusch's Cognitive Ability Scale | Raven's CPM |
|---|---|---|
| Raven's CPM | -.28** | - |
| CHEXI Total Score | .65*** | -.13 |
| Composite Variable of EF | .03 | -.37*** |

*Note.* *p < .05, **p < .01, ***p < .001

A significant relationship was observed between the Waschbusch's General Cognitive Ability Scale and the Raven's CPM, suggesting these two measures are likely to assess similar constructs of cognitive abilities. Interestingly, parent-rated CHEXI was significantly related to parent-rated Waschbusch's General Cognitive Ability Scale, whereas the composite variable of neuropsychological tasks was significantly related to performance-based Raven's CPM. These relationships indicate that the types of measures in similar mode are more likely to correlate than with measures in different mode.

## 3.4 Discussion

### 3.4.1. RRBs across Conditions

The first objective of this chapter was to examine RRB occurrences across three conditions: Task, Preferred and Neutral condition, of children with ASC and typical development, using observational-based and parent-report measures. Relative to TD children, children with ASC have been found to display significantly more RRBs across all three conditions: when they were performing a task, doing something they preferred, and doing something that they neither liked nor disliked. However, during the Neutral condition, the relationship was weak. In general children with ASC display RRBs more frequently than TD children which is not surprising as RRBs are one of the hallmark characteristics in ASC.

Children with ASC particularly displayed significantly more RRBs in the Preferred condition compared to the Neutral and Task conditions, which implies that if RRBs serve as Emotion Regulation mechanism as suggested by Rodgers et al. (2012), children with ASC are more likely to use RRBs to regulate their positive arousals but not so much on negative arousals. As mentioned earlier, six children of ASC walked away, nine got distracted and looked away, and one engaged in Self-Injurious Behaviours when tasks were given. Perhaps children with ASC are more likely to use other behaviours such as walking away, looking away or engaging in potentially harmful behaviours to regulate negative arousals. On the contrary, TD children displayed more RRBs during the Neutral condition than the Task and Preferred conditions. These results suggest that RRBs potentially serve different functions in children with ASC and TD children. For children with ASC, RRBs are likely to serve as a regulatory mechanism when they are excited and when they are engaging with activities that aroused them positively. For TD children, from the homeostatic point of view, RRBs seems to potentially counterbalance under stimulation as they are more likely to engage in

RRBs when they are presented with activities they neither liked nor disliked, which may lead to boredom. These findings reject the traditional definition of RRBs as purposeless behaviours (Bodfish et al., 2000; M. Turner, 1999) because these behaviours have been found to serve specific functions.

In Willemsen-Swinkels et al. (1998), children with higher IQs displayed RRBs during the “composure” condition, whereas children with lower IQs were found to use RRBs to regulate their emotions across all conditions (i.e., composure, excitement and distress). However, it is important to note that, all participants in Willemsen-Swinkels et al. (1998) had developmental delay such as ASC, ADHD, and language development. In the current study, we examined cognitive and IQ levels of all children with both Waschbusch’s Parent-Rated General Cognitive Ability Scale and Raven’s CPM, and no significant differences was found in cognitive and IQ across ASC and TD groups, indicating the children from these two groups were at similar cognitive and IQ level. We further explored the relationship between cognitive ability, IQ levels and RRBs but found no significant relationship between cognitive ability and RRBs nor IQ and RRBs, in the total sample, TD and ASC groups. This result implies that contrasting with Willemsen-Swinkels et al. (1998), cognitive skills and IQ may not have a significant effect on RRBs. However, the differences between our findings and Willemsen-Swinkels et al.’s (1998) findings can be due to the different data analysis methods, in which we examined the correlation between IQ and RRBs, whereas they categorised the participants into different IQ groups and examined the differences.

### 3.4.2. Does Emotion Regulation mediate the relationship between Executive Functioning and RRBs?

The second objective in this chapter was to investigate whether Emotion Regulation was the mediator in the relationship between Executive Functioning and RRBs. First, we replicated Chapter 2 by using similar parent-report rating scales to examine these variables. As an extension from Chapter 2, we replaced parent-report scale with neuropsychological tasks to examine Executive Functioning and observational-based measures to examine RRBs. It is believed that examining Executive Functioning and RRBs with different measurements yields a more meaningful interpretation of the roles Executive Functioning and Emotion Regulation play in this relationship.

The pattern of findings in the total sample, TD and ASC groups were similar to the findings from Chapter 2 except the direct effect from Executive Functioning to RRBs in the TD group. The relationships between the study variables were further analysed by examining Executive Functioning with a set of neuropsychological tasks (i.e., Go/No-Go and DCCS) and by observing participants' RRBs during the non-emotional (Neutral) state. It is interesting that the pattern of results changed when we changed the measurement from parent-report rating scales to neuropsychological tasks and observational-based measures. These findings supported the previous studies in which the, arguably, more objective measures may examine different components than rating scales. The indirect effect from Emotion Regulation on RRBs was no longer significant in the total and ASC samples. The only two relationships that remained significant were: 1) general task performance of Executive Functioning → ERC in the total and ASC samples and 2) general task performance of Executive Functioning → RRBs during Neutral state in the total and TD samples. Interestingly, the path from Executive Functioning to Emotion Regulation was not significant in the TD sample indicating that utilizing different neuropsychological measures can likely result in different patterns of relationship. Data were analysed during the Neutral condition because as described previously, little to no intense emotions was postulated during neutral state, in which limited the confounding variables and highlighted the impact of Emotion Regulation skills when there were no intense positive and negative arousals. The relationship between Emotion Regulation and observed RRBs occurrence during the Neutral condition was not significant in the total, TD and ASC groups. However, the direct effect of Executive Functioning on RRBs was significant in the total and TD groups. It is believed that in a situation with little to no intense emotions, Executive Functions had more impact on RRBs than Emotion Regulation.

It is important to mention that the Neutral condition did have the highest incidence of RRBs in the TD group, suggesting that analysing the RRBs only during the Neutral condition to control for valence might not be the most suitable approach when examining the mediating effect of Emotion Regulation in TD children. Apart from observing RRBs without the presentation of any external stimuli, past research had also conducted observation during a variety of daily activity at home setting such as having snack, playing with or without toys, family chores and caregiving (Stronach & Wetherby, 2014). These activities might contain positive emotional valence which can be varied across individuals. Perhaps it is important to identify the degree of emotional valence in each activity while examining the relationship between RRBs and Emotion Regulation.

Interestingly, when we examined the relationship between the parent-report questionnaire, RBQ-2, and the duration percentage of RRBs across all conditions, the results from the RBQ-2 were significantly related to the RRBs during the preferred condition. This finding suggested that parents might pay more attention to their child's RRBs when they engaged in preferred activities and completed the parent-report scale based on this impression. This finding might explain why the patterns of mediation during the employment of rating scales were different from when the duration percentage of RRBs during the Neutral condition were used for analysis.

### 3.4.3. Executive Functioning and Emotion Regulation

The relationship between Executive Functioning and Emotion Regulation was examined in this chapter with neuropsychological tests and questionnaires. The parent-report Emotion Regulation rating scale was significantly related with the parent-report Executive Functioning rating scale as well as the composite variable of Executive Functioning task performance in the total sample. These findings indicated a potential relationship between Emotion Regulation and Executive Functioning which supports the findings from the past studies. Given past research, we could say that different constructs in Executive Functioning relate differentially to Emotion Regulation as neuropsychological and parent-rating measures of Executive Functioning examined different cognitive constructs but the relationship between Executive Functioning and Emotion Regulation remains significant in both measures. Perhaps, given that Emotion Regulation involves a variety of adaptive and maladaptive strategies, different Executive Functioning constructs might be related to different Emotion Regulation strategies. These results were observed in the past studies, for example, working memory and set shifting were found to correlate with Emotion Regulation strategies such as reappraisal (McRae, Jacobs, et al., 2012; Opitz et al., 2014; Schmeichel et al., 2008b) and suppression (Schmeichel et al., 2008b).

The relationship between Executive Functioning and Emotion Regulation was examined whilst controlling for the Waschbusch's Parent-Rated General Cognitive Ability Scale and Raven's CPM in a partial correlation. Interestingly, although performances in Raven's CPM was closely related to neuropsychological measures of Executive Functioning, the relationship between Executive Functioning and Emotion Regulation remained significant. This result indicate that Executive Functioning and IQ are independent entities and the relationship between Executive Functions and Emotion Regulation is not affected by the individual's IQ.

### 3.4.4. The relationship between Parent-Report and Observational measures of RRBs

As mentioned earlier, the parent-reported RRBs using the RBQ-2 were found to positively correlate with observed RRB occurrences when children were engaging in preferred activities, but not when children were performing a task or when their emotions were in a neutral state. To be specific, the reported lower-order RRBs or motor/sensory repetitive behaviours correlated with the observed RRBs when children were performing the task. The higher-order RRBs or rigidity/routines/preoccupation with restricted interests, were significantly correlated with the observed RRBs during preferred activities. The former finding implies that parent-reported lower-order RRBs are more likely to represent their child's behaviours during negative arousal and the latter finding implies that higher-order RRBs reported by parents are more likely to represent their child's behaviours during positive arousal. Taken together, parents tend to report their child's RRBs that are more likely to represent the child's behaviours during positive or negative arousals rather than in a neutral state. This finding is understandable because if RRBs serve as Emotion Regulation mechanisms, these behaviours are more apparent during emotional states than during a neutral state. Due to its significant occurrences during positive and negative arousals, parents are more likely to attend to and notice these behaviours. According to an early study by Chamberlain & Reid (1987), parents are able to provide reliable day-to-day information on behaviours that are salient or problematic (e.g., aggressive and crying behaviours). It is hypothesized that parents reported children's RRBs based on their observation during periods when the behaviours are most salient and frequent, such as during the presence of emotional triggers. It is noteworthy to mention that it was beyond the scope of this chapter to examine the factors of higher-order RRBs with observational measures and only repetitive motor movements (or lower-order RRBs) were defined for behavioural coding. Therefore, with the limited data that are available in this Chapter, it may not be sensible to suggest that higher-order RRBs are relevant to emotions present during the engagement with preferred activities whereas lower-order RRBs are more relevant to emotions during task performance. Future research should investigate whether parental report of higher-order RRBs are correlated with the information gathered using observational measure.

### 3.4.5. The relationship between Parent-Report and Neuropsychological measures of Executive Functioning

The IES score of the DCCS task was found to positively correlate with the parent reported general Executive Functioning, working memory, and response inhibition, in the total sample. However, the Go/No-Go performance was not significantly correlated to the parent reported general Executive Functioning nor its specific constructs such as working memory and response inhibition. These specific findings indicated that the DCCS neuropsychological task could still measure particular Executive Functioning constructs that were measured by parent-report rating scales, CHEXI, which contrasted with the findings from the past research (Anderson et al., 2002). On the other hand, the IES of the Go/No-Go task did not correlate to the parent report results perhaps due to the inappropriate difficulty levels for TD group as most of the participants from this group were able to complete the task with 100% accuracy within a short duration. In general, the findings highlight the importance of utilising neuropsychological tasks at appropriate difficulty levels to better measure the Executive Functioning constructs.

### 3.4.6. Limitations

The sample size of this study was calculated as a priori based on Chapter 2 but post-hoc Monte Carlo power analysis showed the mediation path from Executive Functioning → Emotion Regulation → RRBs, was lower but still acceptable (.76) for a total sample of 114. The mediation path yielded much lower power of .36 when the results of neuropsychological tasks were used to represent the general Executive Functioning for analysis. Perhaps, the lower power was because there was shared variance from the reporting methods that gave the impression of relationships that are driven by these design artefacts rather than genuine, robust, relationships.

Mediation analysis method can also be another reason resulted in low power. Mediation can be tested using several methods such as structural equation modelling (SEM; Cole & Maxwell, 2003), bootstrapping (Mackinnon et al., 2004) and regression-based tests (Hayes & Rockwood, 2017). Although regression-based mediation analysis is the most preferred and most frequently used by researchers (Fritz & Mackinnon, 2007), concerns have been raised over its statistical power. The statistical power of a hypothesis test refers to the probability of detecting an effect in its presence (Neyman & Pearson, 1933). Statistical power ranges from 0 to 1 and conventionally, a power at .80 and above is considered adequate in Psychology research. Mackinnon et al. (2004) reported lowest power in mediation analysis when the traditional z test and jackknife were used, followed by the second group of measurements such as the percentile bootstrap, bootstrap-t, and Monte Carlo tests. The third group of tests which consist of M test, Empirical-M test, and the bootstrap-Q, have more power and test that has the highest power was bias-corrected bootstrap. Monte Carlo power analysis has comparable power but may not be the best test for mediation analysis, which can affect the power analysis in this chapter.

It is worth mentioning again that 19 children from the TD group were attending remedial classes for additional academic support in subjects such as English, Bahasa Malaysia (BM), Mandarin and Mathematics, namely TD_R. The data analysis was conducted by grouping TD and TD_R groups together in one group as there was no significant difference in the mean score between TD and TD_R groups in the four main domains. However, based on our findings, there was also no significant difference between TD_R group and ASC group in Executive Functioning (both parent-report and neuropsychological measures) as well as Emotion Regulation. The lower power in the mediation path in this study can also be resulted from the characteristics of participants in this study. The low power in this study might indicate that Emotion Regulation might be mediating the relationship between Executive Functioning and RRBs to certain extent of degree when neuropsychological and observational measures were used.

Another limitation of this study is the missing frequency data of repetitive behaviours. Repetitive behaviours were measured only with duration in this chapter as the measurement of every single occurrence of the rapid repetitive movements was not possible as the onset and offset of each occurrence can be challenging to identify and determine. Comprehensive definitions of repetitive behaviours were created to reduce the impact of the absence of frequency data. Additionally, repetitive movements that involve other body parts besides arms, hands, and fingers were not identifiable because participants were in a seated position with lower limbs obscured from the camera in all conditions, which potentially led to an underestimation of repetitive movements observed during the session. It is also believed that technology which enables automated detection and measurement of repetitive movements provides a new and promising methodology for studies in RRBs, which will be explored in Chapter 4.

Both TD and ASC groups showed ceiling effects in the Go/No-Go task, and TD experienced moderate ceiling effects in the DCCS task. The impact of ceiling effect was reduced by utilizing the inverse efficiency score (IES) of the neuropsychological tasks for analyses rather than performance accuracy or reaction time solely. Moreover, the use of parent-report measures used in this Chapter can also counteract the ceiling effect by providing information representing individuals' performances in the real-world. Perhaps the future studies can consider the appropriateness of classical neuropsychological tasks if it involves both TD and ASC samples. Researchers can prepare an additional set of neuropsychological tasks with higher difficulty level for participants who score more than 90% in the original task. Alternatively, a few strategies have been suggested to counteract and prevent ceiling effects which include but are not limited to 1) employing the reaction time instead of the performance accuracy for analysis (Sikström et al., 2016), 2) incorporating a range of differing difficulty levels in a task (Dodell-Feder et al., 2013), and 3) use stimuli that are representative of real-life social interaction (Dodell-Feder et al., 2013). In the current study, the reaction time between TD and ASC groups were not significant and therefore, we used the IES to counteract the ceiling effects.

### 3.4.7. Summary

This chapter addressed the limitations of the previous chapter by utilizing neuropsychological tasks and observational-based methods together with parent-report measures to answer four research questions. Firstly, children with ASC displayed significantly more repetitive behaviours in all three conditions relative to TD children. Children with ASC displayed significantly more RRBs in the Preferred condition, whereas TD children displayed more RRBs during the Neutral condition than the Task and Preferred conditions. These findings imply that RRBs may serve different functions in children with

ASC and TD children, in which RRBs are likely to serve as a regulatory function for children with ASC in an exciting environment. On the other hand, RRBs seem to potentially serve as adaptive behaviours which occur in response to under-stimulation for TD children when they were presented with activities neither they liked nor disliked. Secondly, similar to the findings from Chapter 2, Emotion Regulation mediated the relationship between Executive Functioning and RRBs in the total sample and the ASC sample, when rating scales were used. However, when rating scales were replaced with neuropsychological tasks and observational-based methods, Emotion Regulation no longer had a mediating effect. These findings could be driven by the possibility of shared underlying response biases in parents' impressions, in which parents might have expectations that RRBs are related to Emotion Regulation. Meanwhile, neuropsychological tasks may not be as sensitive as parental report in examining the overall Executive Functioning and therefore potentially undermines the findings. We know that Executive Functioning and Emotion Regulation were found to be significantly correlated in the total sample regardless of whether Executive Functioning was measured with ratings or neuropsychological measures. This relationship remains significant when IQ was controlled. The relationship between Executive Functioning and Emotion Regulation was significant in almost all mediation analyses. These findings imply that RRBs are likely to serve as a regulatory mechanism for children with ASC during positive emotional valence whereas for TD children, RRBs seems to serve as adaptive behaviours when they are under stimulation.

# 4 Chapter Four: Measuring Repetitive Behaviours with Automated Technology

## 4.1 Introduction

Studies on ASC have increased rapidly over the past decade which reflects the growing prevalence rates from an estimated 1 in 88 in 2008 (Centers for Disease Control and Prevention, 2012) to 1 in 44 in 2018 (Maenner et al., 2021). The prevalence of RRBs particularly repetitive motor movements can be as high as 88% in children with ASC (Chebli et al., 2016; Grossi et al., 2021). Despite the high prevalence of RRBs reported in individuals with ASC, the majority of studies of ASC focused on social and communication deficits (Lewis & Bodfish, 1998), perhaps due to the lack of standardised measurements. A variety of tools have been developed to measure RRBs, including traditional methods such as paper-and-pencil rating scales, direct observation, and video-based coding. The last decade of research has brought new and promising measurement techniques which facilitate automated sensing and measuring. The discrepancy of methodology and metrics in measuring RRBs might lead to challenges in comparing the same behaviours across different measures, which consequently contribute to the inconsistent conceptualisation of RRBs. The impact of methodological discrepancy might be one of the reasons why contradictory findings were uncovered in Chapter 2 and Chapter 3. In Chapter 2, Emotion Regulation was found playing a mediating role in the relationship between Executive Functioning and Restricted and Repetitive Behaviour (RRBs) in the total sample and the autism spectrum condition (ASC) group when these relationships were examined using rating scales. When this study was replicated in Chapter 3, Emotion Regulation remained a mediator in the total and ASC samples. However, when rating scales were replaced with observational measures in

Chapter 3, Emotion Regulation no longer had mediating effects. In this Chapter, we aim to focus on the development of RRBs measures and we recognise the need of having a suitable RRBs measure which can objectively and automatically identify and detect the variant repetitive behaviours. We recognised the need of having a time-effective, convenient and automated measurement for identifying, detecting and analysing repetitive behaviours in autism. The progression of technology has brought new measuring techniques which facilitate automated pose estimation and movement detection. Although there is still no research using this technology to analyse repetitive behaviours in autism, we believe this is a promising beginning for researchers in autism studies.

**4.1.1 Paper-and-Pencil Rating Scales**

Paper-and-Pencil rating scales usually involve informants providing global impressions of the frequency, duration, or severity of the RRBs based on their general observations. For self-report rating scales, the participants are the informants themselves, providing information regarding their own behaviours. In parent-report, teacher-report or caregiver-report rating scales, the individuals' parents, teachers or caregivers are the informants who provide information based on their observations on the individuals' behaviours. This approach might contain either an interview, a questionnaire, or an observation scale. The Autism Diagnostic Interview-Revised (ADI-R; Le Couteur et al., 2003), Autism Observation Scale for Infants (AOSI; Bryson et al., 2008), Communication and Symbolic Behaviour Scales Developmental Profile (CSBS-DP; Wetherby & Prizant, 2002) and Diagnostic Interview for Social Communication Disorders (DISCO; Wing et al., 2002) are classic examples of a semi-structured/structured interview conducted with the parents of individuals with ASC. In addition to items focusing on RRBs, these interviews consist of items focusing on other impairments in the domain of language and communication, and social interactions. For example, the ADI-R is a 93 item semi-structured interview widely used to diagnose ASD and to distinguish it from other developmental disorders. There are three functional domains in the ADI-R: Language and Communication, Reciprocal Social Interaction, and Restricted and Repetitive Behaviours and Interests.

Instruments created for the sole purpose of measuring RRBs include the Repetitive Behaviours Scale-Revised (RBS-R; Lam & Aman, 2007), the Repetitive Behaviour Questionnaire (RBQ; Turner, 1995) and its revised version, the Repetitive Behaviour Questionnaire, 2nd version (RBQ-2; Leekam et al., 2007). Although these measures were created to assess RRBs solely, each of these measures includes and excludes different dimensions of RRBs, and has different number of subscales, making it challenging to draw any conclusions about which of these measures is sensitive enough to capture the broad diversity of RRBs. For example, the RBS-R consists of 43 items divided into six subscales including: stereotyped behaviour, restricted behaviour, routine behaviour, sameness behaviour, compulsive behaviour, and self-injurious behaviour (Lam & Aman, 2007). The RBQ (M. Turner, 1995) consists of 33 items in which 29 of these questions measure four types of behaviours such as repetitive movements, restricted interest, sameness behaviour, and repetitive use of language. A summary of domains or factors and the number of questions in each instrument used to measure RRBs is presented in Table 4.1.

**Table 4.1**

*Summary of instruments used to measure RRBs and their number of questions and domains.*

| RRBs measures | ADI-R | DISCO | SCQ | RBS-R | RBI | RBQ-2 |
|---|---|---|---|---|---|---|
| Total of RRBs Questions | 12 | 47 (Uljarević et al., 2022) | 8 (Lord & Rutter, 2003) | 43 (Mirenda et al., 2010) | 50 (South et al., 2005) | 20 (S. Leekam et al., 2007a) |
| Total Questions | 98 | 320 | 40 | 43 | 50 | 20 |
| Repetitive motor behaviors | - | 7 | - | 6 | 12 | 5 |
| Unusual sensory and object focused interests | - | 14 | - | - | 12 | 4 |
| Sensory sensitivity | - | 6 | - | - | - | - |
| Insistence on sameness | - | 7 | - | 10 | 12 | 7 |
| Circumscribed/restricted interests | - | 7 | - | 4 | - | 7 |
| Stereotyped language | - | 3 | - | - | - | - |

| | | | | | | |
|---|---|---|---|---|---|---|
| Self-injurious behaviour | - | - | - | 8 | - | - |
| Compulsive behaviours | - | - | - | 8 | - | - |
| Ritualistic behaviours | - | - | - | 7 | - | - |
| Restricted and Repetitive Behaviours and Interests | 12 | - | 8 | - | - | - |
| Other Repetitive Behaviours | - | - | - | - | 16 | - |

Abbreviations: ADI-R, Autism Diagnostic Interview-Revised (ADI-R; Le Couteur et al., 2003); DISCO, Diagnostic Interview for Social Communication Disorders (DISCO; Wing et al., 2002); SCQ, Social Communication Questionnaire (SCQ; Berument et al., 1999); RBS-R, Repetitive Behaviours Scale-Revised (RBS-R; Lam & Aman, 2007); RBI, Repetitive Behaviour Interview (RBI; Turner, 1997); RBQ-2, Repetitive Behaviour Questionnaire, 2nd version (RBQ-2; Leekam et al., 2007).

It may be a naïve wish to have a “gold standard” rating scale for measuring RRBs given the complexity of these behaviours. However, undoubtedly paper-and-pencil approaches are one of the most convenient, time-efficient and easy-to-implement measurement methods, especially during extenuating circumstances such as the pandemic. However, from a measurement perspective, rating scales can have questionable accuracy which can derive from the informant's subjective impressions of the behaviours (Johnston & Pennypacker, 2009) and they often fail to capture inter-individual variation in the temporal and topographical dimensions of behaviours (McEntee & Saunders, 1997; Pyles et al., 1997a). Alternatively, researchers have used methods such as direct observation and video-based coding for measuring RRBs.

#### 4.1.2 Direct and Video Observation

Direct observation also involves informants rating a sequence of repetitive and stereotypical behaviours. Yet, this approach focuses on direct observation and recording rather than relying primarily on a general impression. In contrast to the paper-and-pencil rating scale, direct observation yields less subjective but more objective data. There are two types of measurements in direct observation: continuous and discontinuous. Continuous measurement records every occurrence of a specific behaviour during a predetermined period, such as frequency (the ratio of count per observation time) and duration (the total extent of time in which a behaviour occurs). Discontinuous measurement divides an observation period into intervals, with the occurrence of behaviour scored during the interval (Fiske & Delmolino, 2012). Interval recording and momentary time sampling are examples of discontinuous measurement for direct observation.

Restricted and repetitive behaviours could involve multidimensional repetitive movements that occur at high speed. The onset and offset of the occurrences of every single motor movement can be challenging to identify and determine. These barriers make data collection with continuous measurement close to impossible for in-person direct observation (Gardenier et al., 2004). Continuous measurement may also require the observers to count every instance of behaviour that occurs at high rates (e.g., hand flapping and scripting) and record multiple behaviours simultaneously. A high amount of attention and effort is required by the observer which may impact the accuracy of the data collected. Mudford and colleagues (2009) reviewed 93 studies (from 1999 to 2009) that contained continuous measurement and found all studies reported inter-observer agreement data and none reported observer accuracy. Considering the broadly defined characteristics of RRBs, discontinuous measurement may be a more suitable approach as the behaviours appear to occur in episodes.

Pyles et al. (1997) were among the pioneers advocating the use of discontinuous measurement for the data collection of RRBs. They created the Stereotypic Analysis Data Collection Form for the concurrent data collection of environmental variables and behaviours. This form helps researchers determine the differential rates of RRBs and their relationship with environmental events, such as the presence and absence of toys, training materials, and adult's attention. In their study, six 1-minute observations were conducted every 30 minutes, and this 1-minute observation was further broken down into twelve 5-second intervals. The occurrence of the targeted RRBs was scored if it happened anytime during the interval, whereas a non-occurrence was scored if the behaviour did not occur for the entire 5-second interval. This measurement is commonly known as partial interval recording (PIR).

Gardenier et al. (2004) combined discontinuous measurement and video-based methods to compare the accuracy of partial interval recording (PIR) and momentary time sampling (MTS). The most significant advantage of video-based coding is its capability to replay the recorded session and collect offline data to ensure all occurrences have been documented. As indicated previously, PIR focuses on the overall occurrence of behaviour during the interval; meanwhile, MTS records the occurrence of the behaviour only at a specific time such as the last one second of the interval. In the study, 600 observations each lasting 30 minutes were videotaped. Occurrences of behaviour (i.e., stereotypy) were collected with four different measurements: PIR 10-s, MTS 10-s, MTS 20-s and MTS 30-s. Gardenier et al. (2004) calculated the percentage difference between the actual duration of stereotypy and duration collected by each estimate method by dividing the duration differences by the duration measure and multiplying by 100% [(estimate duration – actual duration)/actual duration x 100%]. Across all participants, PIR 10-s was found to overestimate the actual duration by an average of 164%, whereas MTS 10-s, MTS 20-s and MTS 30-s over- and underestimated the actual duration by an average of 12%, 25% and 28%. MTS has smaller error margins compared to PIR in general. Moreover, MTS 10-s is reported to yield the most accurate data across all levels of RRBs and MTS-20s produce the most accurate estimates for moderate-level of RRBs. If these findings are correct, the choice of the interval is as important as the types of measurement. Gardenier et al.'s (2004) study has highlighted the importance of evaluating the measurement methods used in recording RRBs.

A significant advantage of combining discontinuous measurement and video-based methods is that full attention may not be required which make it feasible in applied settings and to collect concurrent data collection of behaviour and environmental variables. However, observers who are skilfully trained to collect both data are essential but scarce in many research and clinical settings. Studies that involve observation intervals often involve multiple observers who usually undergo lengthy training before the experiment to ensure they achieve at least 90% of interobserver agreement (IOA) (Gardenier et al., 2004; Matson & Nebel-Schwalm, 2007; Pyles et al., 1997). For example, the observers might be given two weeks of didactic instruction and in-vivo modelling as well as approximately five hours of direct training in Pyles et al. (1997).

Observation and discontinuous measurement are more likely to generate data with higher accuracy and validity compared to paper-and-pencil rating scales, but unlike continuous measurement, it does not record every possible behavioural occurrence but produces only estimates of the true level of behaviours. This approach poses another challenge for objective measurement. As pointed out earlier, a continuous measurement may not be the most suitable apparatus for measuring RRBs due to its multidimensional and high-speed characteristics. This may be addressed with video-based method which allows the session to be re-played to collect offline data for all occurrences. Then again, this method can be impractical and tedious considering the time consumed in watching the video several times to observe and document the behaviours (Matson & Nebel-Schwalm, 2007).

### 4.1.3 Wearable automated technology

The approaches used to document RRBs so far fail to accurately assess the multidimensional and high-speed movements in objective manners. Thus, there is need for a real-time data collection that could address the high-speed characteristics of RRBs. The rapid growth of machine learning and artificial intelligence encourages researchers to explore the possibility of developing an automated technology to replace traditional measurements for RRBs. Bao and Intille (2004) designed a recognition algorithm to detect 20 daily physical activities using the data acquired from wireless sensors worn on the thigh and wrist. This study has frequently been cited in research focusing on automated sensing technology and the development of smartwatches as a tool for research.

Westeyn et al. (2005) built an automated monitoring system, based on the algorithm developed by Bao & Intille (2004), to detect autistic repetitive movements using the Hidden Markov Model (HMM). HMM is a statistical technique commonly used in machine learning to make inferences and to identify the most appropriate set of outcome sequences given a set of probabilities. In other words, a broad range of data is provided to train and construct several types of HMM (e.g., one for a target movement) and these data are used to detect the specific movement based on the trained model. Westeyn et al. (2005) trained seven models each representing a repetitive movement (i.e., hand flapping, drumming, hand striking, pacing, rocking, spinning, toe walking and miscellaneous). The participants wore a sensor on their wrist, back of their waist and left ankle. Then, the data from the sensors were transmitted to a Personal Server located in the participant's pocket or backpack. The approach from Westeyn et al. (2005) could only achieve 68.57% of accuracy in detecting the seven types of repetitive movements indicated previously. However, they argued that the system does not need 100% accuracy in view of the fact that low false positive is acceptable in applied settings as long as the technology is feasible. This argument would have been more persuasive if the authors had tested the system with individuals with ASC rather than a typically developing adult mimicking stereotypical behaviours.

Building upon Westeyn's study, Albinali et al. (2009) successfully tested the HMM system on six individuals with ASC aged between 12 to 20 years old. A higher percentage of accuracy of 88.6% was observed in their study. The increase in accuracy could be attributed to the different body parts (i.e., left wrist, right wrist and chest) where the sensors were attached. This result was further supported by later studies (e.g., Amiri et al., 2017;

Coronato et al., 2014; Gilchrist et al., 2018; Goodwin et al., 2011; Goodwin et al., 2014; Min & Tewfik, 2011; Mohammadian Rad et al., 2018). Generally speaking, mobile and wearable real-time automated recognition technology may be likely to yield more objective, time-efficient, and precise data compared to other traditional approaches. Although this technology may offer the possibility for a more accurate assessment of RRBs, for both research and practice, they can be impractical, as some participants do not want to wear these sensors and tried to take them off (Goncalves et al., 2012). As mentioned in the previous chapters, it is commonly known that most individuals with ASC have sensory sensitivity issues. While advancing the wearable technology for automatic detection and measurement, researchers have overlooked one aspect that is likely to affect its feasibility and effectiveness.

#### 4.1.4 Video-based Pose Estimation

A novel video-based approach was developed by Kang et al. (2016) using video-based technologies together with automatised detection of body poses, the Visual Gesture Builder (VGB) from Microsoft Kinect, to detect repetitive motor movements. The tool was tested with twelve actors performing three types of repetitive motor movements (i.e., hand flapping, spinning, and body rocking) and three types of non-autistic motor movements. For hand flapping, the tool evaluated the deviation angle between forearm joint and wrist joint. For spinning, the deviation angle between hip and the socket joints was evaluated. Lastly, for body rocking, the tool evaluated the deviation angle between the spine base joint and the spin-shoulder joint. The tool detected the target as body rocking when the deviation angle reached a value of 10 degree or higher (see Kang et al., 2016). However, the study did not operationally define the repetitive motor movements and did not address the variation and diversity of autistic repetitive motor movements. For example, some individuals with

ASC flap their hands by moving their entire arm while having elbow staying bent, with their wrists flicking back and forth. Some individuals with ASC flap their hands by having the entire arm still while waving their hands up and down rapidly. Thus far, no study has provided clear definitions for all types of repetitive motor movements, and it has been claimed to be unrealistic to do so due to their broad diversity. Despite of the difficulties, we created nine operational definitions in Chapter 3 to describe the repetitive movements that involved arm(s), hand(s) or fingers with codes including: “Arm_Only”, “2Arms_Only”, “Arm_Object”, “2Arms_Object”, “Arm_BodyPart”, “2Arms_BodyPart”, “Hands_Fingers_Only”, “Hands_Fingers_Object”, and “Hands_Fingers_BodyPart”. Any other repetitive movements involving other body parts besides arms, hands, and fingers were categorised under the code “Other.” The participants’ behaviours were coded and analysed with the ELAN (Version 6.4) [Computer software, 2022], based on these operational definitions. However, manual behavioural coding and analysis can be very time-consuming, in which you have to first identify and allocate the keypoints on the body parts, draw a line connecting these keypoints and then measure the angle between these lines.

Human 2D pose estimation is a real-time system that can automatically detect human body, face, hand and leg, and then allocate anatomical keypoints on these body parts. Top-down pose estimation first detects humans in the image, forms a bounding box of every detected human, and then predicts human joints within the bounding box. The computational duration of these top-down approaches for each detection is proportional to the number of people in the image: The more people there are, the greater the runtime. Recently, Cao et al. (2017) proposed a human 2D pose estimation algorithm using bottom-up approaches. In contrast to top-down approaches, bottom-up approaches first detect the key points of the human body in the image, then assemble the detected key points and associate them with the corresponding target. These approaches are more attractive than the top-down approaches as they have the potential to decouple the computation duration from the number of people in the image. In the pose estimation tool designed by Cao et al. (2017), a two-branch multi-stage Convolutional Neural Network (CNN) was designed to detect body parts (confidence maps) and associate body parts (part affinity fields) in the image (see Cao et al., 2017 for illustrations). Then bipartite matching was used to associate body parts of humans and finally these parts were assembled into full body poses in the image. Their study demonstrated that this approach is sufficient to produce high quality parses of body poses. To the best of our knowledge, this cutting-edge technology has not been used in autism research to measure autistic repetitive movements.

#### 4.1.5 Objectives

We recognise having a suitable measuring tool for identifying and detecting the variant repetitive behaviours is one of the main criteria to better understanding RRBs. This tool would have to be able to detect human body parts and then localise anatomical keypoints, and recognise specific kinematic patterns, as presented in RRBs. To achieve this, this system must be able to measure the angle between individual body parts and report the frequency, duration, and amplitude of repetitive movements in a precise and reliable manner. Such a system could then be used to measure repetitive motor movements directly in individuals with ASC.

As mentioned previously, we know that pencil and paper rating scales can have questionable accuracy and often fail to capture inter-individual variation, whereas direct and video observation can be impractical and tedious considering the time consumed in watching the video several times to observe and analyse the behaviours. Wearable automated technology might not be the most suitable tool as most of the individuals with

ASC have sensory sensitivity issues and they often refuse to put on the sensors on their body. Cao et al. (2017) introduced one of the first real-time pose estimation systems that can automatically detect human body, face, hand and leg, and then allocate anatomical keypoints on these body parts from video so that participants do not have to wear any sensor. This system is considerably more objective compared to rating scales and can significantly reduce the time consumed due to its automaticity. However, this technology does not generate and collect data automatically for behavioural analysis and has not been used in autism research to measure autistic repetitive movements.

In this chapter, to build a technology that not only can detect body parts and allocate keypoints automatically but also generate and collect behavioural data, we modified the Open Pose (Cao et al., 2017) so that angular data can be generated and collected automatically from this tool, namely OpenPose_Angle. We aim to investigate the precision and sensitivity of this modified software in detecting and measuring repetitive body movements of children with ASC. To achieve this, as the first step, we focused on the estimation of landmarks and the angle of individual body segments using standardized pictures in Study 1. The reproducibility of keypoints and consistency of keypoint placement were examined. In Study 2, the OpenPose_Angle system was used to measure a systematic body movement, Jumping Jacks, performed by typically developing individuals. The angular data generated and collected by the OpenPose_Angle were compared with the angular data that were measured manually with the Kinovea, a free software for 2D movement analysis. Lastly, the OpenPose_Angle system was used to measure random repetitive motor movements displayed by children with ASC extracted from Chapter 3.

## 4.2 Study 1 Introduction: Automated Measurement of Movements generated by PowerPoint Slides

In this thesis, we modified the Open Pose algorithms and added automatised computation and generation of angular data functions in the OpenPose_Angle. In relative to the traditional methods such as questionnaires, interviews, and direct observations in studying autistic behaviours and movements, the main strengths of OpenPose_Angle are its automatised pose estimation, keypoint placement, computation and generation of angular data. However, this technology is still considerably novel with limited data on its consistency, sensitivity, and specificity, we are unsure the degree to which it is suitable to be used in autism research. In Study 1, we aimed to examine the consistency of automatic localisation of keypoints on individual's body segments which is important for the computation and generation of angular data as the data are generated based on these keypoints. As we focused particularly on shoulder and elbow angles only in the current study, we created a 10-second movement or video with a PowerPoint document consisting of ten slides repeating two positions (90 degrees and 180 degrees arm positions). This method was used to ensure the body parts are consistently presented at the specific location for each target arm position to reduce the degree of noise generated by real time pose estimation. On these consistent presentation of arm positions, the OpenPose_Angle estimated and placed keypoints automatically on the neck, left shoulder, left elbow, and left wrist and generated the shoulder and elbow angles. Then, it generated shoulder and elbow angular data and exported them into an excel sheet.

These data were compared to angular data collected through manual keypoints placement and angular computation, which presumably had higher repeatability and precision because based on the past research, the OpenPose might lose some level of detail in keypoint estimation due to factors such as low-resolution images (Groos et al., 2021). Six participants were recruited to place markers at the same four different body parts so that the shoulder and elbow angles could be calculated manually using a movement analysis software, Kinovea. More information regarding the procedures and why Kinovea software was chosen for comparison will be provided in the following section. In Study 1, we hypothesize that the angular data generated and collected from the OpenPose_Angle remain consistent over time. We also hypothesize high agreement between the angular data generated by the OpenPose_Angle and the Kinovea.

## 4.3 Study 1 Method: Automated Measurement of Movements generated by PowerPoint Slides

### 4.3.1 Participants

Participants for this study consisted of six adults between the age of 25 to 50 years, categorised into three groups: non-Expert, Researcher, and Expert. The Non-Expert group consisted of an amateur and an undergraduate student in Psychology who were not familiar with the OpenPose_Angle. The Researcher group consisted of a researcher in developmental psychology and another researcher in movement neuroscience who are not familiar with the OpenPose_Angle but with psychology and movement research. The expert group consisted of the thesis author and a researcher in computer science who were familiar with the OpenPose_Angle coding and system. These participants were recruited based on their areas of expertise to control the effect of individual perception, in which different participants from different areas of expertise may provide distinct estimation of landmark and body keypoint placement.

#### 4.3.1.1 Ethics

This study has been reviewed by the University of Reading Malaysia Research Ethics Committee and has been given a favourable opinion for conduct with *UoRM REC 2019/18* as the unique approval reference number. All experimenters working on this project have had the appropriate criminal records checked by the Ministry of Foreign Affairs and received a Certificate of Good Conduct.

### 4.3.2 Materials and Measures

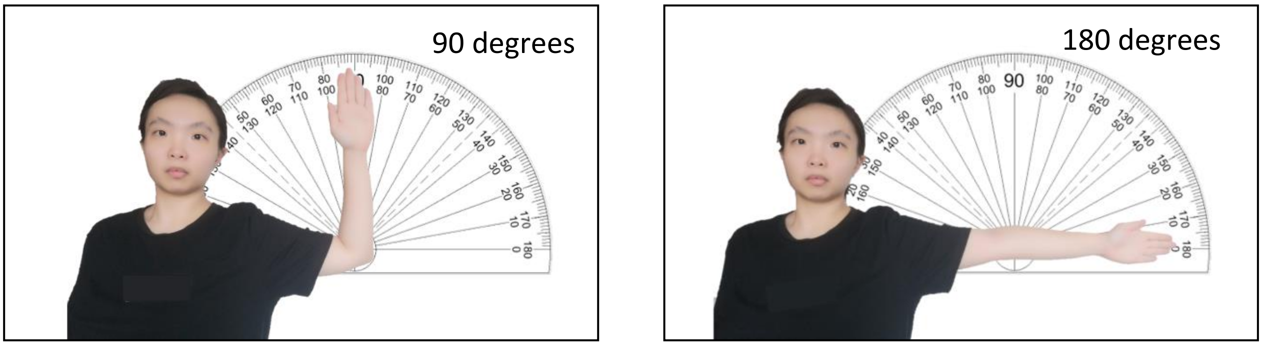
#### 4.3.2.1 Slides displaying 5 cycles of systematic movements

Microsoft PowerPoint slides were used to display changing elbow angles from 90 degrees to 180 degrees. There were two types of slides with one slide displaying the author placing her left arm 90 degrees upward as presented in Figure 4.1 (a) and another displaying the author placing her left arm 180 degrees flat outward as presented in Figure 4.1 (b). These two slides were duplicated into another four sets to form a total of ten slides, showing five cycles of moving elbow when the slide moved from one to the other. A circular protractor was placed in the background to ensure the arm was perceived at 90 degrees and 180 degrees respectively from the eye view.

**Figure 4.1.**

*Sample of arm positions.*

(a) Sample of 90 degrees *arm* position (b) Sample of 180 degrees *arm* position



#### 4.3.2.2 10-second Systematic Movement Video

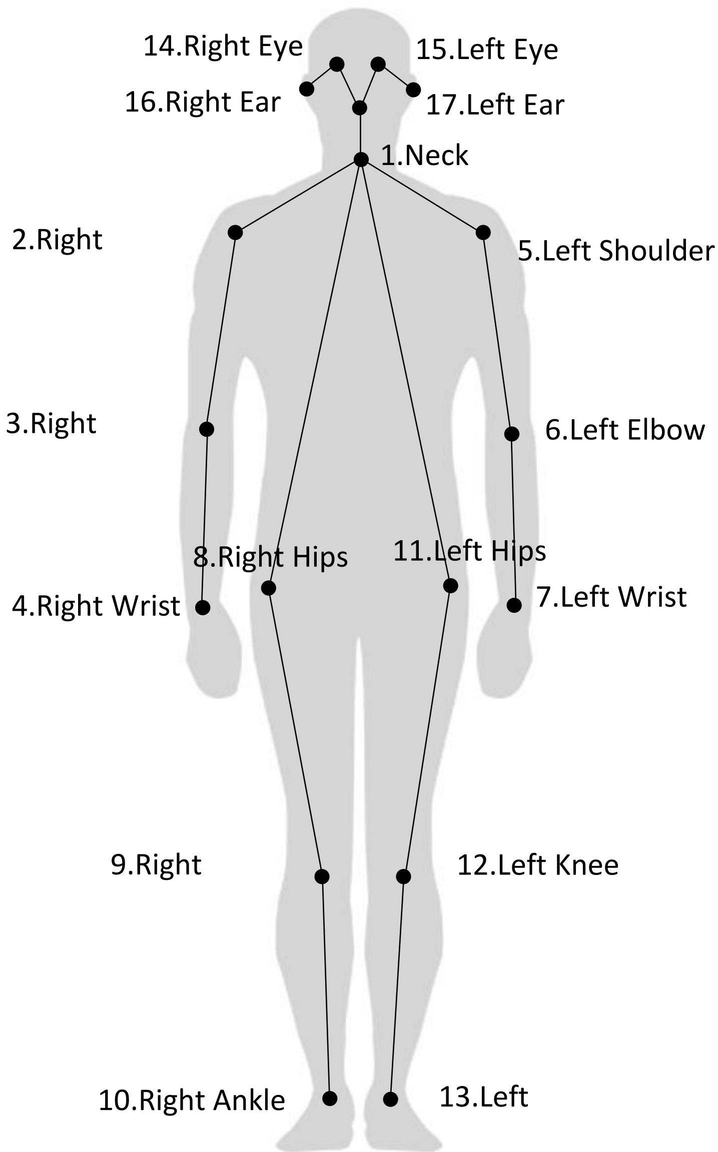
Using Microsoft PowerPoint's *Play Slide Show* command, the slides were presented in full-screen mode. Screen recording began and the slide was moved from one to another with a transition duration pre-set at 1 second, which created a 10-second video. The 10-second video was analysed with a modified OpenPose technology (OpenPose_Angle) and the Kinovea software (version 0.8.15).

#### 4.3.2.3 OpenPose_Angle

OpenPose is a human pose estimation algorithm and a real-time system that automatically detects the human body, face, arm, and foot key points on single images (Cao et al., 2021). Figure 4.2 shows the anatomical keypoints detected by the OpenPose.

**Figure 4.2**

*Anatomical keypoints detected by the OpenPose algorithm*




The algorithm of the OpenPose was available to the public on GitHub and can be implemented using TensorFlow, a free and open-source software library for machine learning. The current study adopted this recently developed technology and added a new algorithm into the system to automatically calculate the angles between shoulder and elbow, and elbow and wrist during the real-time analysis. These angles were measured in radians using the formula, $\theta = \text{atan}(y/x)$. This formula was carried out with a Python math function, math.atan2(), to detect the slope and calculate the angle between the opposite length (y) divide by the adjacent length (x) to the specified point $(x, y) \neq (0, 0)$ in radian. All angles were converted from radians into degrees by multiplying them by 180 and then dividing by the pi value (i.e., radian* 180 / 3.142).

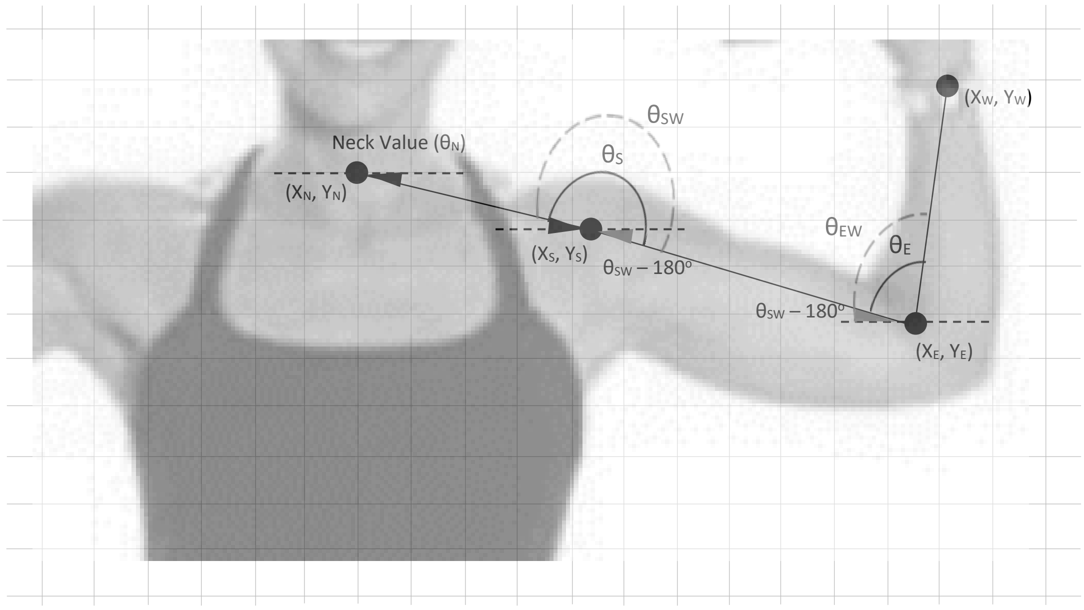
The neck anatomical keypoint was set as the central reference point of all other keypoints. The steps of the mathematical equations to calculate shoulder value and elbow value are as below:

1. First, calculate the neck value ($\theta_N$) with arctan formula, θ = tan-1 (Opposite / Adjacent). The neck value is the angle between the positive x-axis and the line formed between neck coordinate ($X_N$, $Y_N$) and the shoulder coordinate ($X_s$, $Y_s$).
2. To calculate the angle of the shoulder (shoulder value), measure the angle ($\theta_{SW}$) between the positive x-axis and the line formed between the shoulder coordinate ($X_s$, $Y_s$) and the elbow coordinate ($X_E$, $Y_E$). The shoulder value ($\theta_S$) is calculated by using the $\theta_{SW}$ angle to minus the neck value, $\theta_S = \theta_{SW} - \theta_N$.
3. Finally, measure the angle ($\theta_{EW}$) between the positive x-axis and the line formed between the elbow coordinate ($X_E$, $Y_E$) and wrist coordinate ($X_W$, $Y_W$). The elbow value ($\theta_E$) can be calculated using the $\theta_{EW}$ to minus the $\theta_{SW} – 180^\circ$, $\theta_E = \theta_{EW} – (\theta_{SW} – 180^\circ)$.

A schematic illustration of the calculation of shoulder and elbow angles is presented in Figure 4.3.

**Figure 4.3**

*Schematic illustration of the calculation of the angle of shoulder and elbow (in degrees).*

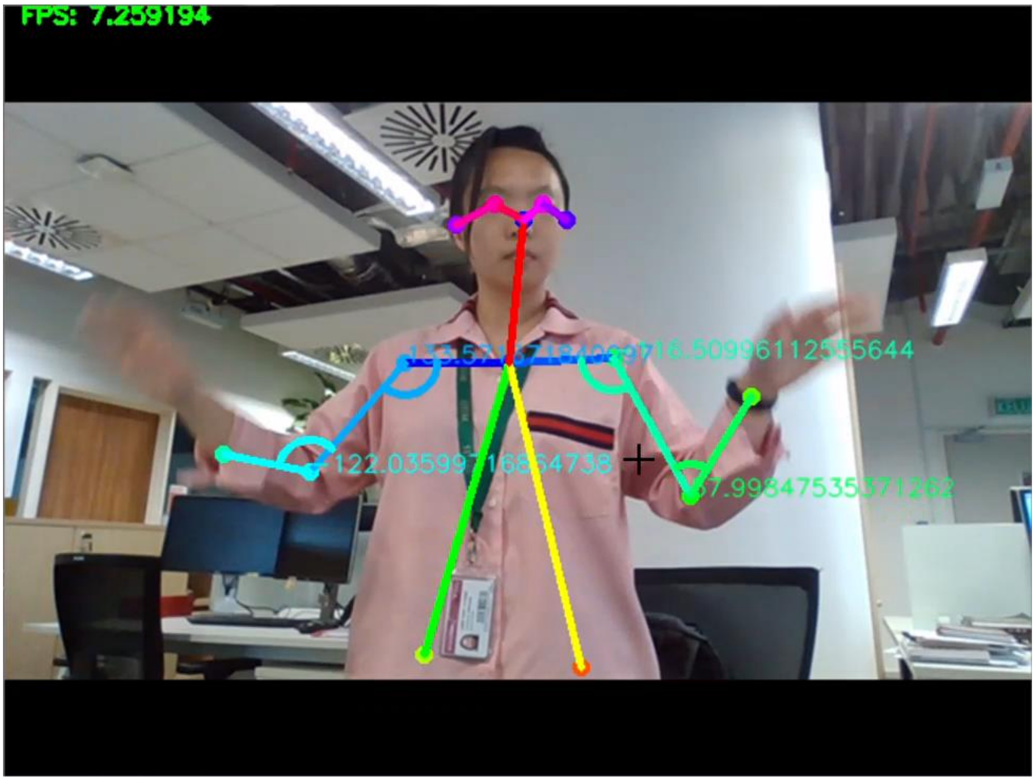



Note. $\theta_N$ refers to neck angle (neck value), $\theta_S$ refers to shoulder angle (shoulder value), $\theta_E$ refers to elbow angle (elbow value), (X, Y) refers to coordinate of a keypoint.

An ellipse, indicating the angle, is drawn on the images during the participant's performance, using cv2.ellipse () method. Besides that, the angle value is written on the images using cv2.putText () method. A screenshot of the OpenPose_Angle environment with ellipse and angular values is presented in Figure 4.4. Another set of algorithms was added so that these data points were compiled and written in an excel sheet automatically following the calculation. The additional algorithm in python language is provided in Appendix 9 and to differentiate the original OpenPose and OpenPose with an additional set of algorithms, the latter was named as OpenPose_Angle.

**Figure 4.4**

*Screenshot of the OpenPose_Angle environment with ellipse and angular values on left and right shoulders and elbows in degree.*




### 4.3.2.4 Kinovea

Kinovea (version 0.8.15) is a free software for 2D movement analysis, frequently used in measuring kinematic parameters. This software provides a set of tools that allows its users to record, rotate, and zoom in for precise measurement and kinematic parameters analysis such as angles, distance and coordinates, frame by frame from a video. Several studies have tested Kinovea software across different populations including autism (Gibbons, 2017; Gómez-Calcerrada et al., 2021) with good results. Gómez-Calcerrada et al. (2021) utilised Kinovea to investigate the walking and climbing gaits and postures of 12 participants with typical development and 12 participants with ASC. Gibbons (2017) used Kinovea to study grasping and eating postures of 19 participants, 9 with ASC and 10 with typical development. Kinovea has been claimed as a valid and reliable motion analytic tool in obtaining angles (Fernández-González et al., 2020; Grigg et al., 2018; Puig-Diví et al., 2019). Although Kinovea requires an experienced experimenter or clinician to use it, it only requires a camera and some markers. Kinovea does not automatically localise anatomical keypoints and calculate angles. User has to place the virtual keypoints over the individuals' body parts, connects the keypoints and draw the angles manually on the images. In this study, four target keypoints (neck, shoulder, elbow and wrist) were placed manually by six participants on each frame of the 10-second video based on the definition given to them. The angles were drawn on the shoulder and elbow, and data were exported onto an excel spreadsheet.

#### 4.3.2.5 Video Analysis Device

The duration for movement analysis with the OpenPose_Angle is determined by the performance of the laptop's operating system and its technical specifications. The researcher used Dell Vostro 5481 with an Intel® Core™ i7-8565 CPU operating at a frequency of 1.80GHz (or 1992 MHz) with 4 cores and 8 logical processors, and an NVIDIA GeForce MX130 Graphics Card. Although NVIDIA GeForce MX130 is a mid-range laptop graphics card, it operates at a frequency of 1109MHz to 1189MHz and its memory is running at a frequency of 1253MHz.

### 4.3.3 Procedure

A PowerPoint document consisting of ten slides with 90 degrees and 180 degrees arm positions was sent to all participants. Participants placed markers (cross shape) at four different body parts including neck, left shoulder, left elbow and left wrist. Sample slides with crosses on the body parts for both 90 degrees and 180 degrees are provided in Figure 4.1. Participants then returned the slides via email.

#### 4.3.4 Data Analysis

Only two angles (shoulder and elbow angles) were targeted in this chapter to investigate the reliability and validity of the automatised measurement, OpenPose_Angle. The duration and speed of the movements was made constant in this study by transitioning from one slide or position (e.g., 90-degree) to the next slide or position (e.g., 180-degree) at 1 second and the total duration of the video or whole movements was 10 seconds. Analyses were conducted comparing the angles extracted from 1) OpenPose_Angle and 2) Kinovea. For OpenPose_Angle analysis, the angle of the shoulder and elbow (in degree) was extracted (see Figure 4.5). For Kinovea analysis, each arm position was added as a new key image, and the angles of the shoulder and elbow were drawn manually with the Kinovea angle tool on each key image. All angular data were then extracted into an excel spreadsheet. A scatterplot comparing angular data between OpenPose_Angle and participants' keypoints placement analysed by the Kinovea was created.

Statistical analyses were conducted using IBM SPSS Statistics for Windows, Version 27.0. As there were more than two raters placing markers on body parts for Kinovea analysis, the intra-class correlation coefficient (ICC) was used to evaluate the reliability (Bartlett & Frost, 2008). ICC values above .90 indicate excellent reliability, values between .75 and .90 indicate good reliability, values between .50 and .75 indicate moderate reliability and values less than .50 indicate poor reliability (Portney & Watkins, 2000).

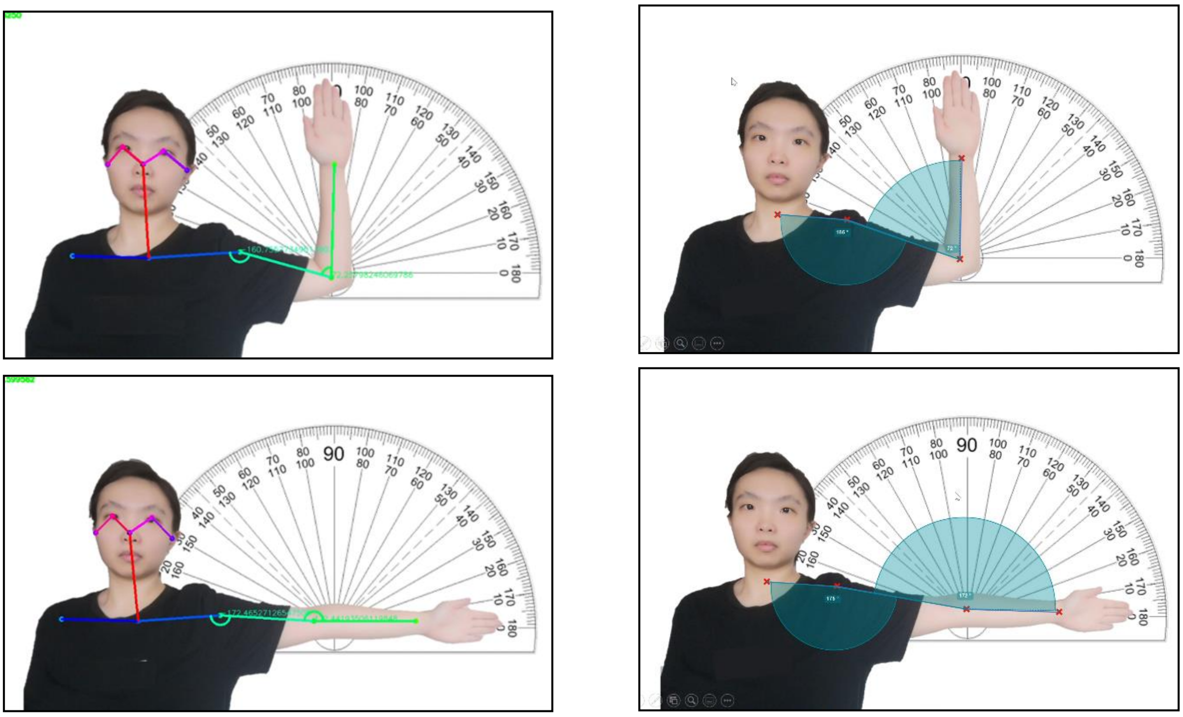
Bland-Altman analysis was conducted to evaluate the agreement between OpenPose_Angle and Kinovea. The bias (mean difference) and the limits of agreement (mean difference ± 1.96 SD, Standard Deviation) are displayed in the Bland-Altman plots.

The mean score is plotted on the x-axis and the mean difference between measurements is plotted on the y-axis. The Bland-Altman plots allow comparison between two measurements and identification of any possible systematic difference or outliers between them (Yeo & Park, 2020).

**Figure 4.5.**

*Screenshot of angular analysis in the OpenPose and Kinovea environments.*

(a) OpenPose_Angle (b) Kinovea



## 4.4 Study 1 Results: Automated Measurement of Movements generated by PowerPoint Slides

### 4.4.1 Descriptive Statistics

Angular data were collected from the OpenPose_Angle and the Kinovea on two elbow positions (90 degrees and 180 degrees). Means and SDs for the elbow angular data generated by the OpenPose_Angle and Kinovea based on the markers placed by six participants throughout the video are presented in Table 4.2.

**Table 4.2.**

*Means and SDs of elbow's angular data generated by the OpenPose_Angle and Kinovea across five trials.*

| Angular Name | 90-degree Elbow position | |
|---|---|---|
| | Mean | SD |
| OpenPose_Angle | 73.82 | 1.43 |
| Non-Expert 1 | 84.56 | 3.26 |
| Non-Expert 2 | 66.66 | .61 |
| Researcher 1 | 73.32 | 1.25 |
| Researcher 2 | 77.13 | 1.11 |
| Expert 1 | 73.80 | 1.79 |
| Expert 2 | 76.06 | .83 |
| **Angular Name** | **180-degree Elbow position** | |
| | Mean | SD |
| OpenPose_Angle | 172.98 | 1.10 |
| Non-Expert 1 | 181.28 | 2.06 |
| Non-Expert 2 | 159.40 | 2.17 |
| Researcher 1 | 171.16 | 1.60 |
| Researcher 2 | 175.30 | 4.02 |
| Expert 1 | 172.00 | 1.87 |
| Expert 2 | 179.74 | 1.85 |

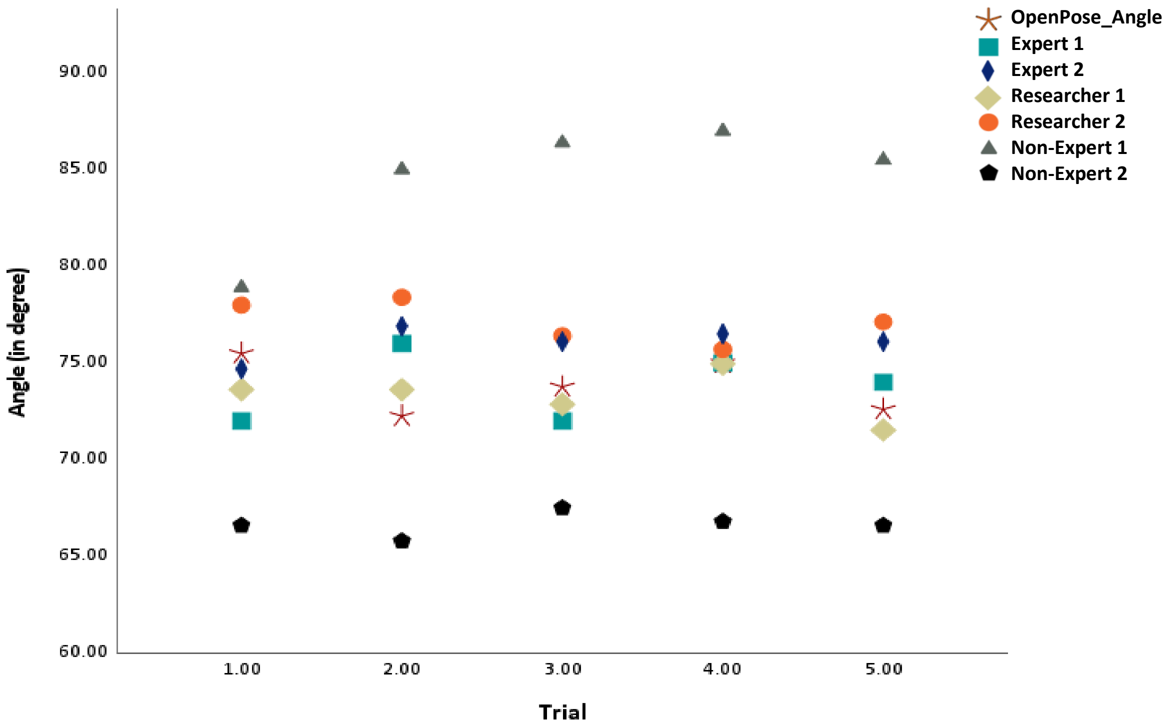
Six participants placed markers manually on four body parts (neck, shoulder, elbow and wrist) for Kinovea analyses. The elbow angular data generated by the OpenPose_Angle when the elbow was in the 90-degree position has a lower standard deviation (*M* = 73.82, *SD* = 1.43) relative to the Kinovea data generated based on markers placed by Non-Expert (*M* = 84.56, *SD* = 3.26, *Min* = 78.90, *Max* = 87.00) and Expert 1 (*M* = 73.80, *SD* = 1.79, *Min* = 72.00, *Max* = 76.00). Meanwhile, the angular data generated by the OpenPose_Angle when the elbow position was at 180 degrees has the lowest standard deviation (*M* = 172.98, *SD* = 1.10) compared to all data generated by the Kinovea. A lower standard deviation (SD) indicates higher precision of the measurement, which refers to the degree of agreement between repeated measurements under the same conditions. A scatterplot comparing the OpenPose_Angle and the Kinovea data of the left elbow angle in the 90-degree position is presented in Figure 4.6.

For the 90-degree elbow position, not one of the angular data displays 90 degrees. Similarly, not one of the angular data displays 180 degrees for the 180-degree elbow position. However, it is important to note that the angular data could be affected by the position of the markers or keypoints. As observed from Figure 4.1, the Non-Expert group formed the highest and lowest value with great discrepancy from other groups for both 90-degree and 180-degree elbow positions. Interestingly, the mean angular data collected from the OpenPose_Angle for both the 90-degree elbow position (*M* = 73.82) and 180-degree elbow position (*M* = 172.98) were the median of each data set.

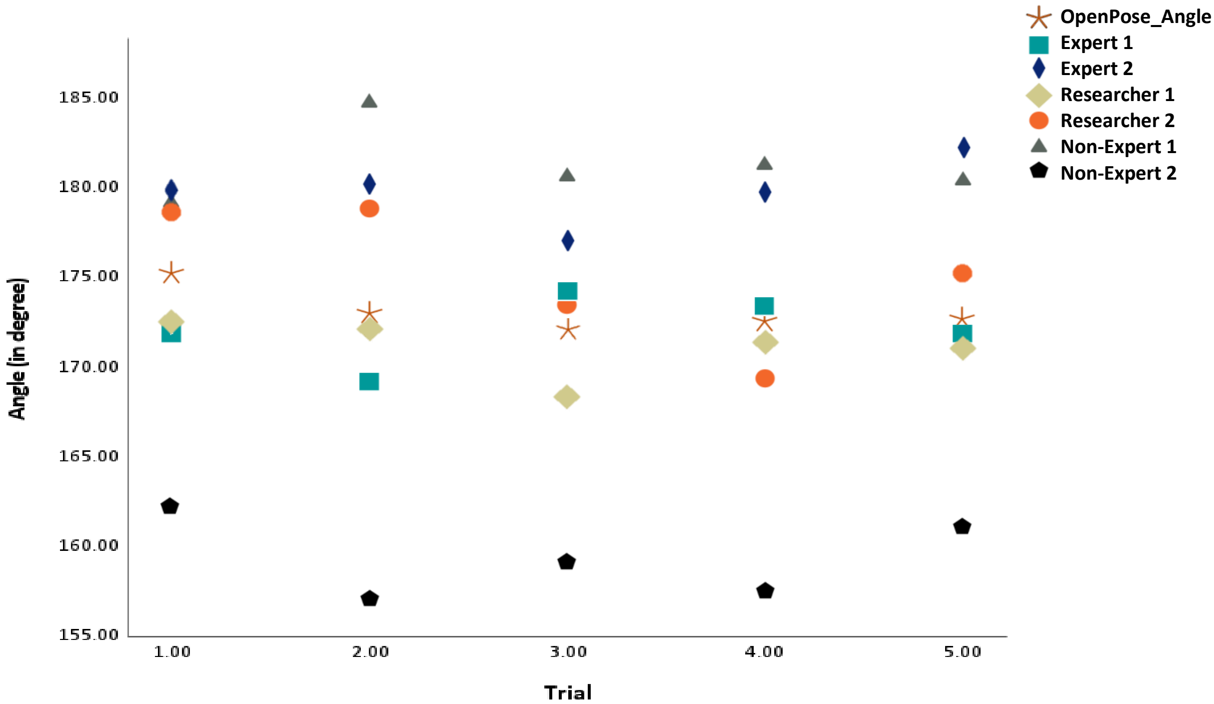
**Figure 4.6.**

*Analyses of left elbow angles by the OpenPose_Angle versus Kinovea (with markers placed by six participants).*

a) *90-degree elbow position*




b) *180-degree elbow position*



### 4.4.2 Agreement

An inter-class correlation coefficient (ICC) was conducted to evaluate the correlation between Kinovea angular data generated based on the markers placed by six participants. The inter-rater reliability reported a correlation for the 90-degree elbow position (ICC = .33, 95% confidence interval [CI] = -1.37, .92, $p$ = .24) and the 180-degree elbow position (ICC = -.06, 95% confidence interval [CI] = -2.74, .88, $p$ = .46).

There are two possible reasons for the negative ICC values. Firstly, the covariance and correlation between Expert 1 and other raters were mainly negative. By removing Expert 1 data from the dataset, the ICC value would be at .47. Secondly, Taylor (2009) stated in his unpublished article that negative estimates of ICC "are possible and can be interpreted as indicating that the true ICC is low, that is, two members were chosen randomly from any class vary almost as much as any two randomly chosen members of the whole population." The negative value may also suggest the variability within participants can be greater than across participants. However, it is important to note that the sample size and the repetitions of trials were low. Although these data should be interpreted cautiously, they provide meaningful information regarding manual rating and perhaps human error.

### 4.4.3 Differences between OpenPose_Angle and Kinovea

There were no significant differences in the average comparison angles between OpenPose_Angle and Kinovea for 90-degree elbow position ($p$ = .09) and 180-degree elbow position ($p$ = .80). Mean differences between systems for 90-degree and 180-degree elbow angles were -1.43 and -.17, respectively. Bland-Altman plots were used to display the relationship between OpenPose_Angle and Kinovea for both 90-degree and 180-degree elbow position. When points scattered above and below zero in a Bland-Altman plot, it suggests that there is no consistent bias of one approach versus the other. In the Bland-

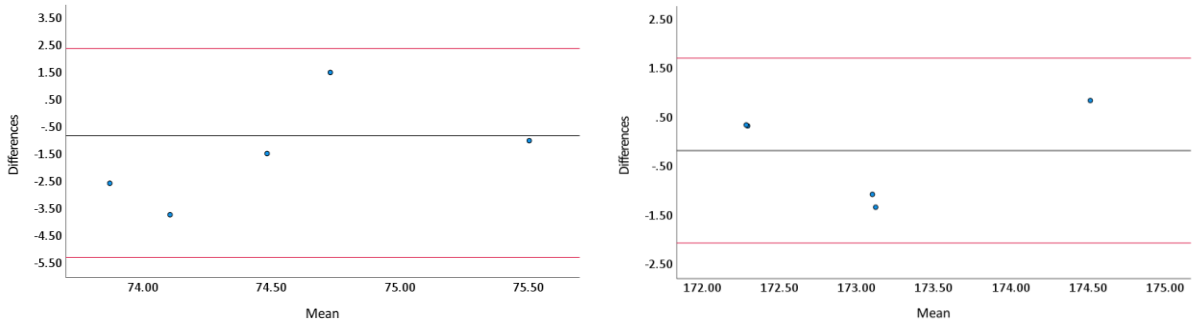
Altman plots, the 95% limits of agreement for 90-degree and 180-degree elbow angles were -5.26 to 2.40, and -2.05 to 1.72, respectively. The Bland-Altman plots for 90-degree and 180-degree elbow angles are presented in Figure 4.7.

**Figure 4.7.**

*Bland-Altman plots comparing results between systems (OpenPose_Angle and Kinovea) for angular data of (a) 90-degree and (b) 180-degree elbow position. Bias (mean difference) [black line] and 95% limits of agreement (mean difference ± 1.96SD) [red lines] are displayed for each position.*

a) 90-degree elbow position

b) 180-degree elbow position




## 4.5 Study 1 Discussion: Automated Measurement of Movements generated by PowerPoint Slides

In this study, we first examined the consistency and precision of the angular data generated and collected from the OpenPose_Angle and the Kinovea. Our results found that the OpenPose_Angle had the lowest SD for the 180-degree arm position, indicating that this measurement had the highest precision for this angular position and was able to place keypoints at similar locations repeatedly under the same conditions. Despite that the precision of measurements that relied on manual keypoint placement was relatively lower, there were no significant differences between the average angles gathered using the Kinovea and OpenPose_Angle. The agreement among the angular data analysed by the

Kinovea based on the markers placed by different raters was low and negative ICC values suggested greater variability within participants than across participants. Higher SDs were found in the Non-Expert group, and then Researcher group, relative to the Expert group and the OpenPose_Angle, suggesting low reproducibility of measurements.

For the 90-degree arm position, not one of the angular data analysed by the OpenPose_Angle and Kinovea presented 90-degree elbow angular data. Similarly, not one of the angular data analysed by these two measurements presented 180-degree elbow angular data. Angles derived from the eye view can be different from angles generated and collected by the OpenPose_Angle and Kinovea depending on the keypoints placed on body parts. Interestingly, the angular data collected based on the keypoints placed by both participants from the Non-Expert group had the highest and lowest mean for both 90-degree and 180-degree arm positions, which also had the highest SD from the total mean. Traditional movement analysis systems involve placing markers on the participant and tracking the trajectories of these markers, such as the optical capture system. Although there are some existing rules for placing markers such as markers being placed at a location that can be identified easily to ensure the reproducibility of the measurements (Chèze, 2014), there is still no ideal method to position the markers. Based on the current findings, human eye view varies which can affect the marker/keypoint placement, and expertise leads to less variability. Besides that, for a 10-second movement, we took an average of 8 minutes and 23 seconds to compute the shoulder and elbow angles and extract the data into an excel sheet, whereas only an average of 1 minute 36 seconds were required to achieve the same tasks using the OpenPose_Angle. Although the latter duration can be affected by the laptop's specifications. Based on the above findings, the OpenPose_Angle is likely to achieve what Kinovea can achieve but with a shorter time, a lesser discrepancy between keypoint locations and consistent reproducibility.

## 4.6 Study 2 Introduction: Automated Measurement of Systematic and Repetitive Movements

The findings from Study 1 indicated that the OpenPose_Angle was able to consistently place keypoint, compute and collect the angular data within a shorter time, compared to the traditional movement analysis systems, in which keypoints and angles were placed and computed manually. However, it is important to note that the movements that were used for analysis in Study 1 were generated by repeating two positions (i.e., 90-degree and 180-degree arm positions) using a PowerPoint document. This method was used to ensure the OpenPose_Angle was able to consistently place keypoints and compute angles without missing details resulted from factors such as low-resolution images or shutter vibration. In Study 2, to reduce the consistency of movement presentation at specific location over time and increase the degree of uncertainty and movement noise, the technology and software were used to analyse a systematic and repetitive movement, Jumping Jacks. First, we conducted a kinematic analysis on these movements using the OpenPose_Angle and compared the results to the data produced by the Kinovea. Then, we examined the sensitivity and specificity of the OpenPose_Angle by comparing its angular data with the frequency of Jumping Jacks cycle counted manually through observation from the Kinovea analysis.

## 4.7 Study 2 Method: Automated Measurement of Systematic and Repetitive Movements

### 4.7.1 Participants

#### 4.7.1.1 Characteristics of the Participants

Eleven typically developing participants, six males and five females, from four age groups were recruited:

1. four children who were 5, 6, and 8 years old,
2. three adolescences who were 11, 12, and 15 years old,
3. two younger adults who were 28 and 30 years old, and
4. two older adults who were 60 and 71 years old.

There was at least one female participant and one male participant in each age group. Participants included in the study do not have any atypical development diagnosis that might affect their motor skills, such as dyspraxia, autism, or any other physical disabilities that prevent them from performing the jumping movements.

#### 4.7.1.2 Recruitment Procedures

Participants were recruited via the university's researcher panels and an email was sent to all staff from the University of Reading Malaysia (UoRM). The potential participants who expressed interest were contacted via phone and screened for eligibility. Participants who met the criteria were provided with the background and procedural information of the experiment during the same contact. Due to the impact of the Covid-19 pandemic, the participants were given a choice to record three videos of their Jumping Jacks by themselves or have the experimenter record their movements in person. Six participants chose to be recorded in person and the five participants who chose to record the movements themselves were provided guidelines for the recording requirements (Appendix 10).

### 4.7.2 Materials and Measures

#### 4.7.2.1 OpenPose_Angle

The OpenPose_Angle, as described in the previous study, was used in this study to obtain angles automatically from Jumping Jacks videos. As some Jumping Jacks videos were recorded horizontally, these videos were rotated by a set of new algorithms. These additional algorithms are presented in Appendix 9.

#### 4.7.2.2 Kinovea

Kinovea (version 0.8.15), similar to the previous analysis, was also used in this study. Key images were created in the Kinovea environment for angular analysis. The first key image was created when the arms are placed overhead before returning to the starting point, and the following key image was created when the arms are resting on the thighs. These cycles were repeated for the rest of the videos.

#### 4.7.2.3 Video Analysis Device

The duration for movement analysis with the OpenPose_Angle is determined by the performance of the laptop's operating system and its technical specifications. We used the same laptop as fully described in the previous study.

### 4.7.3 Procedure

Participants from the onsite recording group were given a copy of an information sheet and a consent form at the beginning of the session. They were informed that their participation was voluntary, and that they could withdraw at any moment. Participants below 18 years old were given an information sheet with pictures to foster their understanding of the procedures and informed consent from their parents. Participants from the self-recording group received an email consisting of the information sheet, consent form, step-by-step guidelines for their recordings and Jumping Jacks, and an access link to the One-Drive folder where they could upload the videos upon completion. The step-by-step guidelines came with pictures to enhance their understanding, especially for the younger participants. The guidelines are presented in Appendix 11.

All participants were required to perform Jumping Jacks for three trials and each trial lasted for 30 seconds. Jumping Jacks were chosen as they involve a set of similar repetitive movements, similar to repetitive behaviours displayed by people with autism but more systematic. One cycle of the Jumping Jack movement began with the participants having their legs together and arms resting on their thighs. Then, the participants jumped with their legs spread a few centimetres wide and the arms came above the head moving towards each other, which might follow with a clap. Finally, their arms returned to the starting position with the feet closing together and the arms back to the thighs. A schematic illustration of one cycle of Jumping Jacks is shown in Figure 4.6. The participants were allowed to take a break for as long as they wanted in between trials.

**Figure 4.8**

A schematic illustration of one cycle of Jumping Jacks.

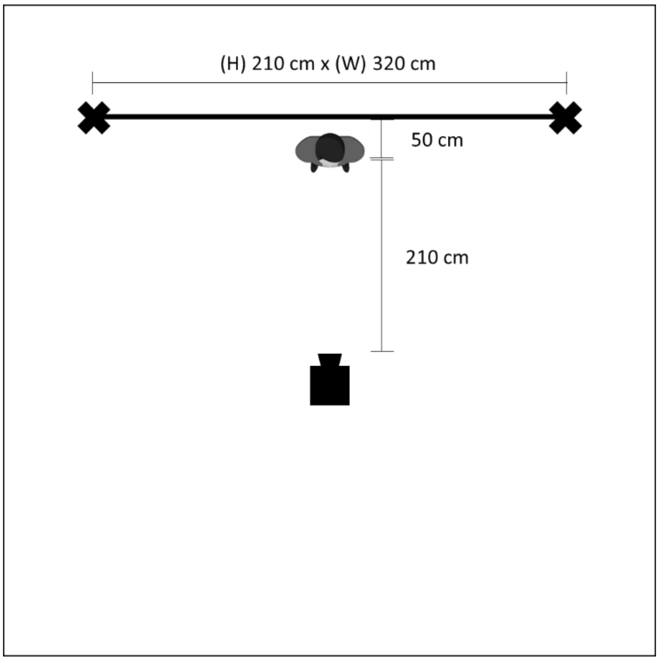
For participants who recorded their own Jumping Jacks, they were required to place their phone on a tripod and ensure the camera captured their or their child's entire body when they were jumping. They were also given an information sheet for securely uploading their videos onto the University's secured folder (see Appendix 12)

#### 4.7.3.1 Set-up

For onsite recording, a plain background was set up with a white background cloth (height = 210 cm, width = 320 cm) to provide clear images and to reduce image noise which might be resulted from random variations of brightness or colour information. The participant performed Jumping Jacks approximately 50 cm in front of the plain background and 210 cm away from the camera (see Figure 4.8). These distances between the participant, background and camera were set so that the participant's movements and all body parts could be captured within the frame while jumping.

**Figure 4.9**

*Experiment set-up overview.*



### 4.7.4 Data Analysis

The OpenPose_Angle was used to analyse Jumping Jacks and the data obtained from this technology was then compared to the data obtained by Kinovea (version 0.8.15). The duration of the video is constant (30 seconds) but the speed of each cycle of Jumping Jack movements varied. The shoulder and elbow angles were drawn and extracted manually with Kinovea software, whereas these angles were extracted automatically from the OpenPose_Angle into a Microsoft Excel sheet. Screenshots of OpenPose_Angle and Kinovea analysis on a child performing Jumping Jacks are presented in Figure 4.9. The sensitivity and specificity of the OpenPose_Angle were measured by comparing its data with observed frequency that was counted manually by the author based on the Kinovea data. The formula for sensitivity and specificity is as below:

$$Sensitivity = \frac{True\ Positives}{True\ Positives + False\ Negatives}$$

$$Specificity = \frac{True\ Negatives}{True\ Negatives + False\ Positives}$$

For these analyses,

1. True Positive refers to the number of times when the smallest shoulder angular data (when both palms were touching one another above head, hereby known as "hit" trial) in between the two largest angular data that were detected by the OpenPose_Angle and the author,
2. False Positive refers to the number of times when the smallest shoulder angular data in between the two largest angular data that were detected by the OpenPose_Angle only but not the author,

3. False Negative refers to the number of times when the largest shoulder angular data (when both arms were resting on thighs, hereby known as “no-hit” trial) in between the two smallest angular data that were detected by the author only but not the OpenPose_Angle,
4. True Negative refers to the number of times when the largest shoulder angular data in between the two smallest angular data were detected by both OpenPose_Angle and the author.

**Figure 4.10**

*Screenshots of OpenPose_Angle and Kinovea analysis on a child performing Jumping Jacks.*


A line graph is generated comparing the angular data obtained from the OpenPose_Angle and Kinovea. The number of Jumping Jacks was first calculated automatically using Python library *scipy.signal.find_peaks()* function to detect where the peaks are located.

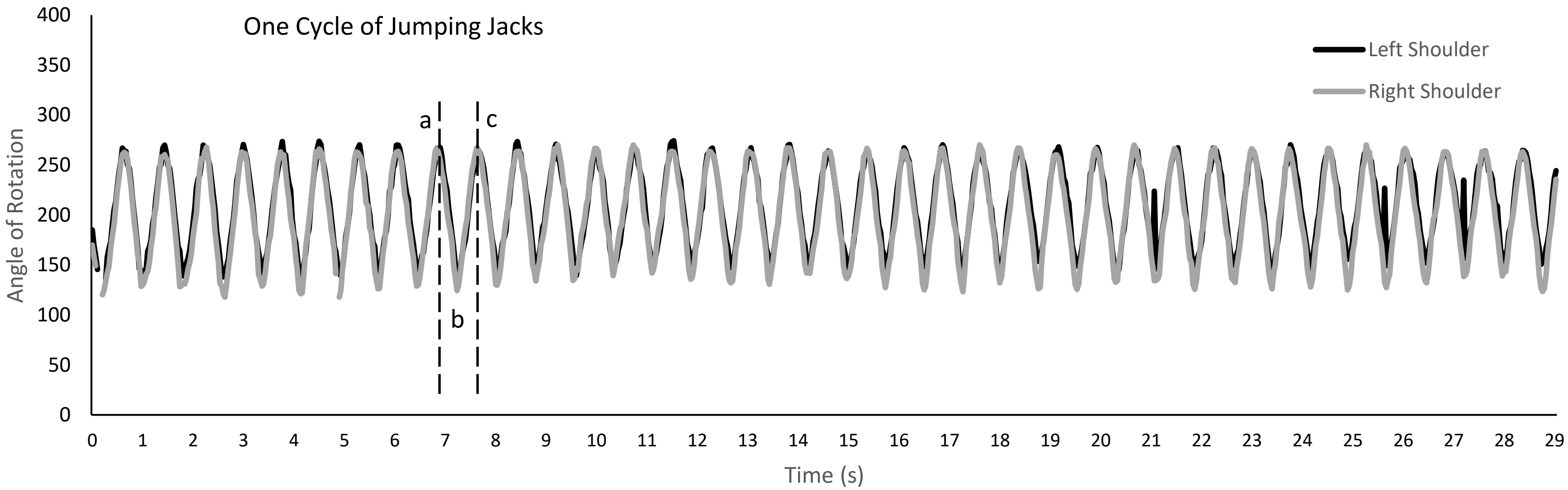
## 4.8 Study 2 Results: Automated Measurement of Systematic and Repetitive Movements

### 4.8.1 Kinematic Analysis

Angular data were generated automatically by the OpenPose_Angle automatically and an example of the kinematic analysis by the OpenPose_Angle is depicted in Figure 4.4. A cycle of Jumping Jacks was defined as moving from resting arms on thighs (largest shoulder angles as shown in Figure 4.4, point a) to having both arms above head touching each other (smallest shoulder angles as shown in Figure 4.10, point b) and finally back to resting arms on thighs (as shown in Figure 4.10, point c). The frequency of Jumping Jack cycles can be extracted directly from the graph.

**Figure 4.11**

*Amplitudes of Left and Right Shoulder Movements decoded by OpenPose_Angle during 30-second Jumping Jacks.*

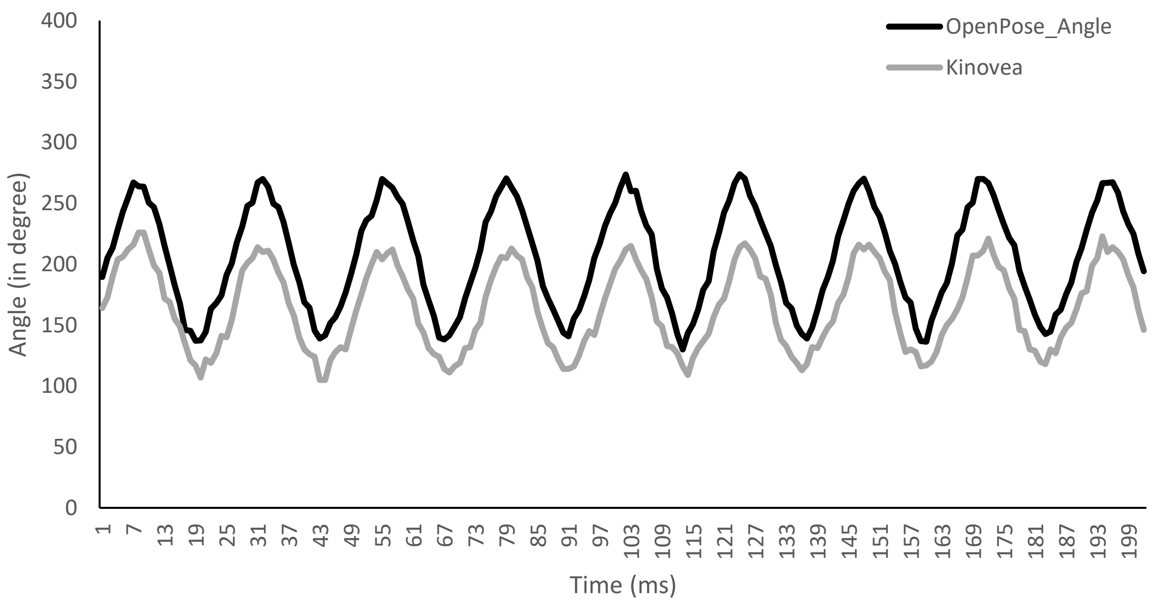



*Note.* This figure shows the angles of the left and right shoulder during the 30 seconds of Jumping Jacks produced by OpenPose. The frequency of the participants' jumping jacks can be extracted from the movement cycle.  Point "a" indicates the beginning of a cycle of Jumping Jacks when arms are down and resting on the thighs. Point "b" indicates the peak of the cycle indicates when arms are lifted above the head. Point "c" indicates the end of that cycle of jumping jacks having both arms returning to the starting point, which also indicates the beginning of the next cycle of jumping jacks.

The comparison of the OpenPose_Angle and Kinovea amplitude graph for the first 200 milliseconds (ms) is illustrated in Figure 4.11. As observed from the graph, shoulder angles generated by the OpenPose_Angle were larger (max = 273.87, min = 130.00, range = 143.86) than angles generated by the Kinovea (max = 273.87, min = 105.00, range = 121.00).

**Figure 4.12**

*Comparison of Amplitudes of Shoulder Movements decoded by OpenPose_Angle and Kinovea.*




The sensitivity and specificity of the OpenPose_Angle were measured by comparing its frequency with the observed frequency. Table 4.3 reported the number of Jumping Jacks cycles for 30 seconds trial based on observation and angular data generated by the OpenPose_Angle.

**Table 4.3.**

*Number of Jumping Jacks during 30-second trial*

| Group | Observed Frequency (Mean) | OpenPose_Angle Frequency (Mean) | Sensitivity | Specificity |
|---|---|---|---|---|
| Child | 29.67 | 30.33 | 1.00 | .98 |
| Teenager | 38.25 | 38.25 | 1.00 | 1.00 |
| Young Adult | 34.5 | 34.5 | 1.00 | 1.00 |
| Elderly | 30 | 30 | 1.00 | 1.00 |

A sensitivity of 1.00 was reported for all groups, suggesting that 100% of the “hit” observed trials were detected by the OpenPose_Angle. A specificity of .98 was reported for the Child group and 1.00 for other groups, suggesting that 2% of the “no-hit” observed trials were detected as “hit” by the OpenPose_Angle for the Child group.

## 4.9 Study 2 Discussion: Automated Measurement of Systematic and Repetitive Movements

In Study 2, we compared the kinematic results of Jumping Jacks gathered by the OpenPose_Angle and the Kinovea, and then we examined the sensitivity and specificity of the OpenPose_Angle. One cycle of Jumping Jacks has been defined as movements beginning from arms resting on the thighs to arms being lifted above the head and back to the starting point. The number of Jumping Jacks from the 30-second recording can be easily counted based on the amplitude graph generated based on the angular data from the OpenPose_Angle. Although the angular data generated by the OpenPose_Angle were relatively larger than the angles measured manually by the Kinovea, the amplitude and movement patterns remained the same. It is important to note that the data provided by

Kinovea displayed a smoother line on graphs than the OpenPose_Angle and thus, this indicates that the OpenPose_Angle might provide more information in relative to the Kinovea although both systems analysed the movements every 0.04 seconds. However, we noticed that the angular data generated by the OpenPose_Angle consisted of up to 12 decimal places whereas the Kinovea provided only whole numbers. Due to the additional information, the automatic peak detection of the cycles might also be easier for the OpenPose_Angle.

The frequency of Jumping Jacks counted based on observation was compared with the frequency counted from the amplitude graph generated by the OpenPose_Angle. The sensitivity of the OpenPose_Angle was reportedly high for all age groups but the specificity in the Child group was lower than in the Teenage, Young Adult and Elderly groups. Pose estimation on children has been a huge challenge in Deep Learning architectures due to insufficient datasets on children's movement to enhance the performances of the existing algorithms (Farinella et al., 2017). Although it might seem that children and adults have the same body shape, they are not the miniatures of adults. The development of body parts occurs continuously but sporadically and not uniformly from birth through old age based on a predictable trend (Huelke, 1998). The automatic estimation of pose in children is therefore a challenge for the research community.

## 4.10 Study 3 Introduction: Automated Measurement of Real-Life Restricted and Repetitive Behaviours

From Study 2, we learnt that the OpenPose_Angle provided relatively more details in angular data and the cycles of repetitive movements can be detected and counted directly from the amplitude graphs. The OpenPose_Angle also had a high sensitivity for all age groups. Although the specificity for the Child group was lower compared to the Teenage, Young Adult and Elderly groups, it was still considerably high. Based on the findings that the OpenPose_Angle can consistently measure real-life systematic and repetitive movements performed by typically developing children, teenagers, adults and elderlies, we used it to measure the spontaneous and random repetitive behaviours displayed by children with ASC and TD children.

Study 3 is part of the main study described in Chapter 3, in which participants' repetitive behaviours were analysed and compared across three conditions: Task, Neutral and Preferred. A total of 114 children were recruited and their behaviours were recorded during all conditions. The ELAN (Version 6.4) [Computer software, 2022] was used to code the participants' repetitive behaviours based on the predetermined operational definition (see Chapter 3, p. 144). Only videos that involved apparent arm movements were selected for the OpenPose_Angle analysis because as mentioned previously, we focused particularly on shoulder and elbow angles for this thesis and therefore, the system was design specifically to detect and measure these two angles only. We identified a total of 274 videos with these screening criteria. These videos were analysed by the OpenPose_Angle but only 110 complete datasets were generated from these videos. The factors of missing data will be discussed in the following sections. These 110 datasets were then categorized into four behavioural groups: Clapping, Tapping Object, Hand Swinging, and Jumping & Rocking. For exploratory purposes, an amplitude graph was created for each behavioural group.

## 4.11 Study 3 Method: Automated Measurement of Real-Life Restricted and Repetitive Behaviours

### 4.11.1 Participants

The sample of this study was the same as in Chapter 3. There are a total of 114 children aged between 5 years to 13 years, of which 60 of them are children with typical development and 54 of them had a clinical diagnosis of Autism Spectrum Conditions (ASC).

### 4.11.2 Materials and Measures

#### 4.11.2.1 Recording Device

The sessions were recorded with the experimenter's smartphone, Huawei P20. Huawei P20 records with 4000x3000 pixels at 30 Hertz (Hz) or frames per second (FPS) RGB (Red, Green, Blue) camera with phase detection autofocus (PDAF), contrast autofocus (CAF), laser and depth autofocus software. It also has a 20 megapixels monochrome sensor.

#### 4.11.2.2 Video Editing Software

Participants' behaviours were recorded throughout the session, and thus, the recordings during the Task condition included the phase when instructions were given and when participants were taking a break in between. Therefore, the recording parts when the participants performing the tasks were clipped manually using a simple video editing software with a free trial version, BeeCut (Wangxu Technology (HK) Co., 2022).

#### 4.11.2.3 Behaviour Coding Software

As described in Chapter 3, the ELAN (Version 6.4) [Computer software, 2022], a computer software that provides an annotation function for video recordings (Lausberg & Sloetjes, 2009) was used to manually code participants' repetitive behaviours. A controlled vocabulary was created and added to the list of behavioural codes. This template was standardized and used for all video analyses.

#### 4.11.2.4 Video Clipping Algorithm

Annotated parts of the recordings were clipped from the ELAN software using FFmpeg program (Tomar, 2006) for OpenPose analysis. FFmpeg program is a video and audio converter that can be freely downloaded from www.ffmpeg.org. A command script *"\ffmpeg.exe" -i $in_file -vf scale=960:540 -ss $begin(sec.ms) -t $duration(sec.ms) $out_file"* was added to a text file called "clip-media.txt" that was stored in the ELAN installation folder. However, the videos could not be clipped altogether with FFmpeg. Target videos were screened and clipped one at a time manually. Participants who were either displaying repetitive behaviours that did not involve apparent arm movements (e.g., hand movements, leg movements, rocking, finger movements) or not facing the camera throughout the sessions. Apparent arm movements were required for this analysis as the OpenPose_Angle was designed specifically to identify shoulder and elbow angles and the participants were supposed to sit throughout the sessions while completing the activities. These screening criteria identified a total of 274 videos. These videos were analysed using the modified OpenPose algorithm. A new python algorithm was added so that all 274 videos could be analysed automatically (see Chapter 3 for more information).

#### 4.11.2.5 OpenPose_Angle

The OpenPose_Angle algorithm was used in this study to obtain angles automatically from videos across three conditions (Task, Neutral and Preferred). These conditions were described in Chapter 3.

#### 4.11.2.6 Data Compilation

The angular data generated by the OpenPose_Angle algorithm was compiled with Pandas library (The pandas development team, 2020) into a Microsoft Excel sheet. Pandas is an open-source software library built for the Python programming language for data manipulation and analysis.

#### 4.11.2.7 Plot

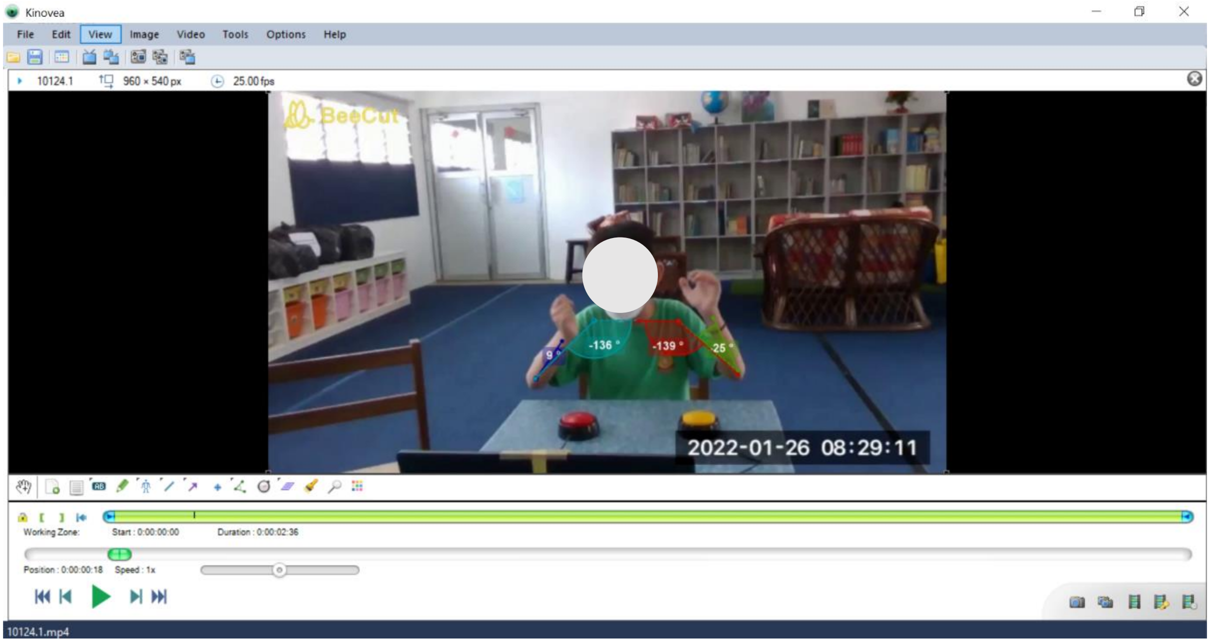
Matplotlib (Hunter, 2007), a 2D graphics library for the Python programming language, was used to create plots based on the angular data produced by the OpenPose_Angle algorithm. Plots generated by the Matplotlib enabled researchers to identify missing data easily and only plots with no missing data were selected for Kinovea analyses.

#### 4.11.2.8 Kinovea

The Kinovea (version 0.9.05) software was also used in this study. Key images were created in the Kinovea environment by placing one at the onset of each RRBs cycle and another at the offset of the cycle. For instance, the onset of a cycle of Hand Flapping movements was described as placing hands furthest apart from each other and the offset was when the surface of both hands touched each other. The shoulder and elbow angles were drawn and named on the first key image. The semi-automated tracking function in the Kinovea was used to gather the angles of movements. To do so, markers were placed on specific body parts (neck, shoulder, elbow and wrist) and when the video was played, the tracking function follows the trajectories of these markers and formed moving angles on the video. The marker position was adjusted when it was dislocated from the target body part during the path creation. A screenshot with angles drawn on recording in the Kinovea environment was shown in Figure 4.12.

**Figure 4.13.**

*Angular analysis of a child's repetitive behaviours in the Kinovea environment.*

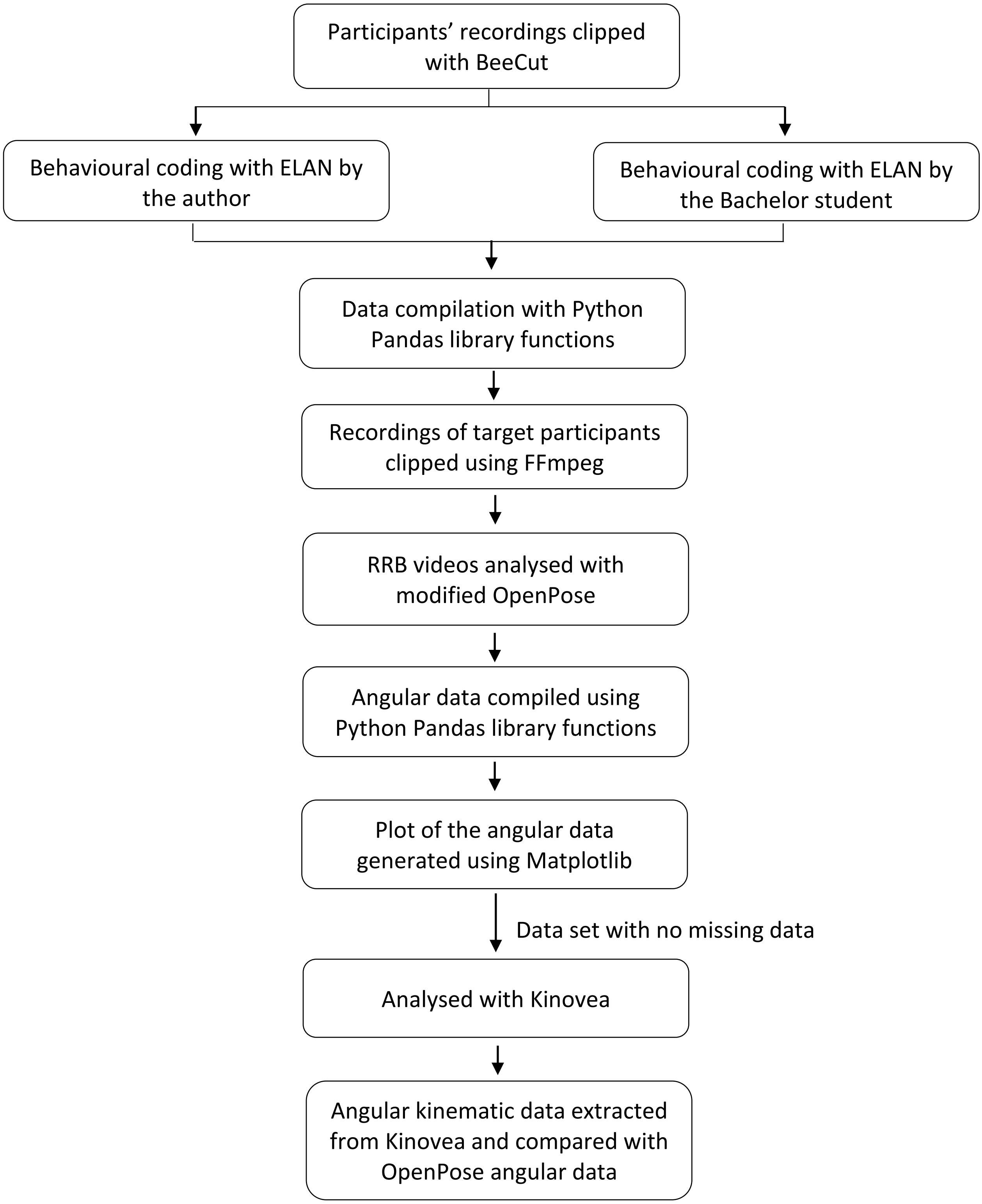



### 4.11.3 Procedure

The recruitment and experimental procedures of this study were described in Chapter 3. Behavioural coding was conducted by the author of this thesis and a student who was doing her Bachelor's degree in Psychology. A schematic representation of the video analysis procedure is shown in Figure 4.13.

**Figure 4.14**

*Schematic representation of the video analysis procedures.*



#### 4.11.4 Data Analysis

The angular data gathered from the OpenPose_Angle and Kinovea were compared. The angular data of all cycles of repetitive movements were graphed automatically using Matplotlib. A total of 263 charts were generated and each chart has four lines representing the angular data of the Left Shoulder, Left Elbow, Right Shoulder, and Right Elbow. Datasets with missing data were eliminated due to their incomplete data. Then, the data were further analysed and repetitive movements that did not involve only arm movements were removed. The remaining angular data, a total of 110 datasets, were compared with the data generated by the Kinovea.

##### 4.11.4.1 Standardization

Repetitive behaviour durations (in percentage) were standardized and converted into z-scores for comparison and analyses across conditions. See Chapter 3 for further description.

### 4.12 Study 3 Results: Automated Measurement of Real-Life Restricted and Repetitive Behaviours

As mentioned in the Method, a total of 110 datasets which presented complete data of repetitive behaviours involving arm were gathered for analysis. Of these 110 complete datasets, 54.55% comprised Left Shoulder angular data, 28.18% Right Shoulder angular data, 11.82% Left Elbow angular data, and 5.45% Right Elbow angular data, suggesting that shoulder angular analyses were more sensitive than elbow angular analyses, particularly Left Shoulder. These complete data can be categorized into four groups: Clapping (50%), Tapping Object (22.73%), Hand Swinging (19.09%), and Jumping & Rocking (8.18%). The definition of these behavioural groups is created based on our observation as presented in Table 4.4.

**Table 4.4.**

Operation definition of behavioural groups with complete data from the OpenPose_Angle analyses.

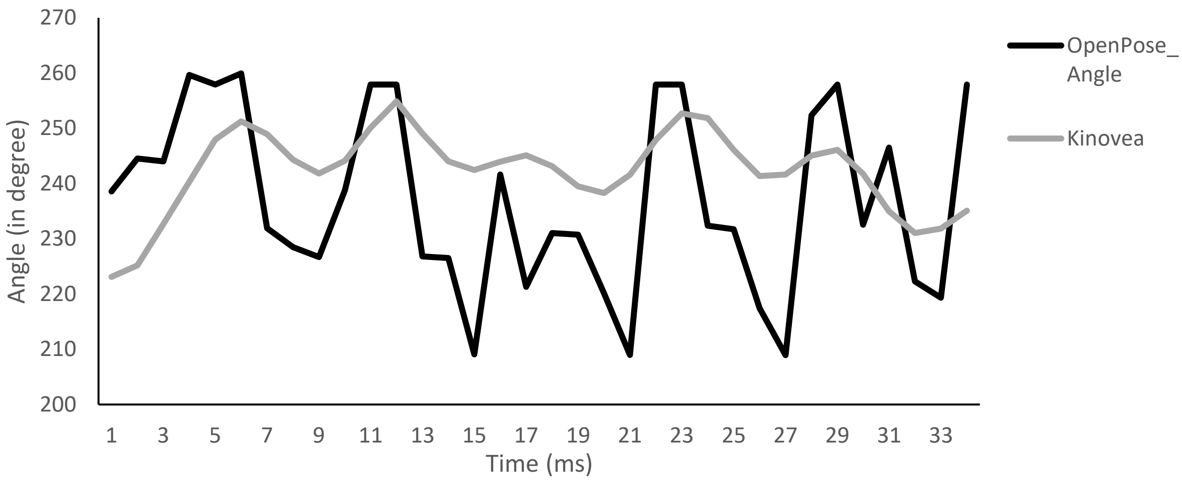
| Behavioural Group | Definition |
|---|---|
| Clapping | A non-contextual behaviour involving one hand touching any surface of the other hand in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles |
| Tapping Object | A non-contextual behaviour involving one or both hand touching the surface of any non-living thing in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles |
| Hand Swinging | A non-contextual behaviour involving one or both arms moving in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles |
| Jumping & Rocking | A non-contextual behaviour involving whole body moving in any direction repeatedly for 2 or more cycles, with no more than 1 second between cycles. |

A comparison of the behavioural pattern generated by the OpenPose_Angle and Kinovea for each behavioural group is presented in Figure 4.14.

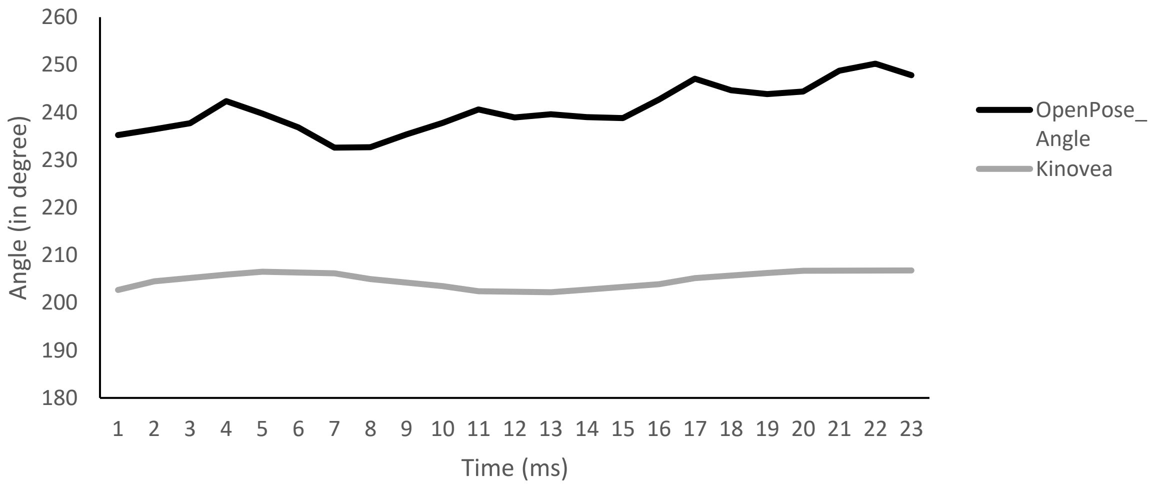
**Figure 4.15**

*A comparison between the OpenPose_Angle and Kinovea of a trial example of different behavioural patterns* based on left shoulder angular data.

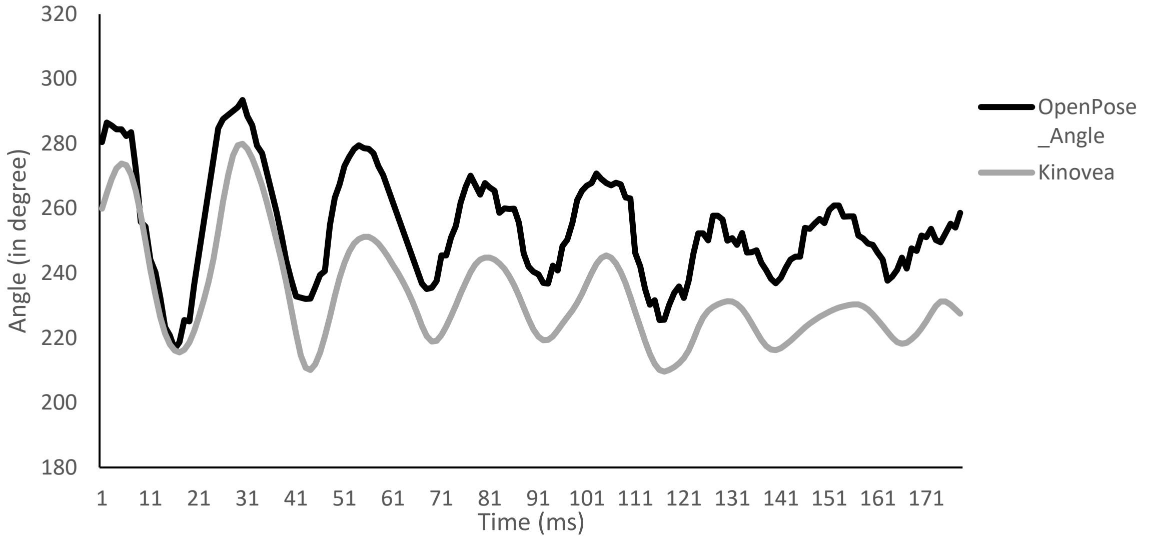
a) Clapping behavioural pattern based on left shoulder angular data



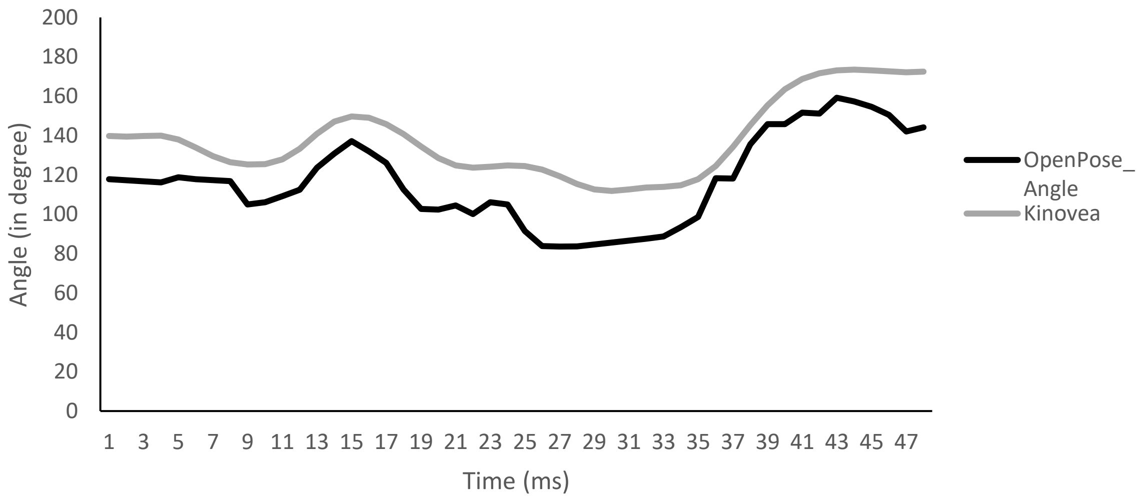

b) Tapping object behaviour pattern



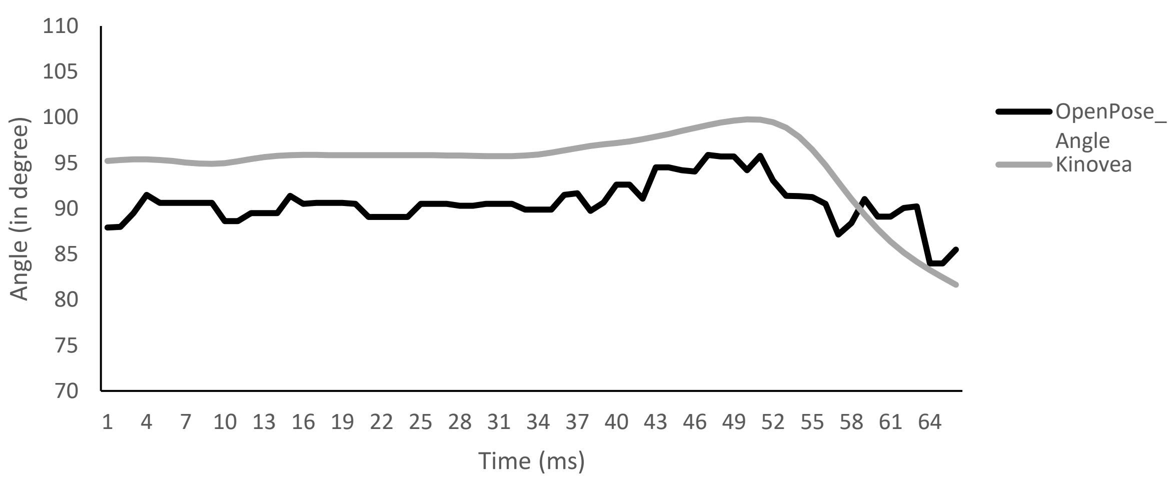
c) Hand Swinging behavioural pattern




d) Jumping & Rocking




e) Non-repetitive behaviours



## 4.13 Study 3 Discussion: Automated Measurement of Real-Life Restricted and Repetitive Behaviours

In Study 3, the OpenPose_Angle was utilised to automatically measure and collect angular data for spontaneous and random repetitive movements. The OpenPose_Angle was only able to collect approximately 10% of complete data from these spontaneous repetitive movements. More than half of these data were Left Shoulder angular data followed by Right Shoulder angular data. These findings can be explained by three reasons. The OpenPose_Angle relies on participants' face to form keypoints for other body parts and therefore when the participants were not facing straight ahead, the camera could not capture the participants' face and the system failed to measure and collect angular data. From a digital imaging perspective, participants' movements were recorded using a phone RGB (Red, Green, Blue) camera with 4000x3000 pixels at 30 Hz. It is believed that higher pixel size can increase sensitivity and improve the accuracy of feature measurements in digital imaging. From a mechanism perspective, shoulders have three degrees of rotational freedom as they can rotate in any direction, whereas elbows can only bend in one direction, giving them one degree of rotational freedom. It is reasonable that movements that involve more apparent angular changes resulting in more complete data. It is important to note that, this study is the first to measure angles automatically using pose estimation algorithms and more studies are needed for more comprehensive interpretation of these findings.

For exploratory purposes, an amplitude graph was created for each behavioural groups with complete data: Clapping, Tapping Object, Hand Swinging, Jumping & Rocking and non-repetitive behaviours, based on the Left Shoulder angular data. The cycles of repetition were apparent on the Clapping and Hand Swinging amplitude graphs only. These findings suggested that automated measurements such as the OpenPose_Angle, are more likely to generate meaningful data for Clapping and Hand Swing repetitive behaviours, demonstrating that these common RRBs could be adequately captured by automated pose capture technologies for future studies.

## 4.14 General Discussion

In this chapter, we modified the OpenPose and developed the OpenPose_Angle that can automatically place keypoints on individual's body segments, compute shoulder and elbow angles, gather and export the angular data onto an excel sheet. Three mini studies were conducted to examine the usability of this system. In Study 1, we examined the consistency of pose estimation and keypoint placements of the OpenPose_Angle over time using a PowerPoint document repeating two arm positions for 10 seconds, generating systematic and repetitive movements with consistent presentation of arm location. Compared to the data collected from Kinovea, in which keypoints were manually placed by six participants, we found the OpenPose_Angle had a high degree of agreement between repeat measures especially for the 180-degree arm position. The OpenPose_Angle took a shorter time, which was only an average of 1 minute 36 seconds to automatically place keypoints, compute the angles, and extract the data into an excel sheet, whereas these steps had to be conducted manually with Kinovea, which took an average of 8 minutes 23 seconds.

After learning that the OpenPose_Angle is likely to achieve what Kinovea can achieve but within a shorter time, a lesser discrepancy between keypoint locations and consistent reproducibility, in Study 2, we used the OpenPose_Angle to measure Jumping Jacks performed by typically developing individuals. We found that although the angular data gathered from the OpenPose_Angle were larger than the data gathered from the Kinovea, the amplitude and movement patterns remained the same. The OpenPose_Angle provided more details than Kinovea which resulted in clearer peak and cycles on the amplitude graphs. The sensitivity of the OpenPose_Angle was also high for all age groups (Child, Teenager, Young Adult and Elderly). Although the specificity for the Child group was relatively lower compared to other groups, it was still considerably high.

In Study 3, as part of the main study described in Chapter 3, the OpenPose_Angle was used to measure the spontaneous and random repetitive behaviours displayed by children with ASC and TD children. Only approximately 10% of complete data were collected from the OpenPose_Angle analysis and more than half of these data were Left Shoulder angular data followed by Right Shoulder angular data. Amplitude graphs were generated for four behavioural groups and the cycles of repetition were apparent on these graphs for Clapping and Hand Swinging beahviours, suggesting that these common RRBs could be adequately captured by the OpenPose_Angle for future studies.

#### 4.14.1 Limitations

Traditionally, a motion capture system using synchronised cameras was usually used for capturing participants' movement from all direction and markers were placed manually on the participants' body for kinematic analysis. However, kinematic analysis using this system can be expensive and time-consuming. In this chapter, automated pose estimation and angular analysis have been shown to be effective even when using low-cost equipment in natural environments. It is important to note that, we focused only on repetitive behaviours involving apparent arm movement. For full-body repetitive movement to be collected, more advance equipment might be needed. The OpenPose_Angle also failed to capture and analyse the participants' behaviours when they were not facing the camera. We believe that providing clear written or verbal instructions reminding the participants to face the camera might be effective for future studies.

Angular data and angular momentum are frequently used in kinematic gait analysis especially in sport science. However, this is the first study that analysed and collected angular data of repetitive behaviours displayed by children with ASC. From the current findings, we have learnt that the amplitude graphs generated based on the angular data gathered from the OpenPose_Angle enabled us to automatically analyse the frequency and duration of behavioural repetitions quickly and easily, which can be important for behavioural analysis. In additional to the frequency and duration, the OpenPose_Angle also provide other information such as the speed and velocity of the repetitive behaviours. Nevertheless, only four behavioural groups were observed in Study 3 and due to the limited amount of complete data, the patterns of these four behaviours were not consistent especially for Tapping Objects and Jumping & Rocking. It is believed that by addressing the limitations of the system and capture more complete data, the OpenPose_Angle can help to analyse autistic repetitive behaviours and their behavioural pattern automatically for behavioural analysis purposes.

### 4.14.2 Summary

Studies have found that individuals with ASC display atypical kinematic movements relative to typically developing individuals (Cook et al., 2013). However, many studies are still relying on traditional methods such as questionnaires, interviews, and direct observations to study autistic behaviours and movements. This chapter provided novel findings regarding automated measurement by modifying a recently developed pose estimation technology, OpenPose, into a system that can automatically measure shoulder and elbow angles, OpenPose_Angle. Three studies were conducted to examine the usability of the OpenPose_Angle. Compared to having participants manually place markers/keypoints on target body parts and analysing the angles using the Kinovea, Study 1 found that the angular data produced by the OpenPose_Angle had a higher precision under the same conditions in general. Study 2 further reported that the sensitivity was reportedly high for all age groups, but the specificity was relatively lower in the Child group. This result was understandable given that it is still a challenge in pose estimation technology in tracking children's movements due to insufficient datasets and images depicting children in pose estimation research. In the last study, only 10% of the total analyses were complete, indicating that although the OpenPose_Angle has high precision and reproducibility of keypoints, as well as high sensitivity and acceptable specificity, this system is still not ready for measuring spontaneous repetitive movements displayed by children with ASC. It is believed that increasing the datasets and images of children's movements can increase the detectability and precision of OpenPose for measuring children's repetitive behaviours. OpenPose utilised deep neural networks, a type of machine learning model, to perform keypoint detection and pose estimation. Deep learning techniques are employed to automatically learn hierarchical representations of features from input images. To train the neural network, large datasets with annotated examples of human poses were used. The network learns to associate input images with the corresponding keypoint locations through a process known as supervised learning. Therefore, having a larger datasets for children's movements can further train the neural network and increase the accuracy and predictability in tacking children's movements.

By addressing the above limitations, it is believed that the OpenPose_Angle can be a useful measurement in not only measuring angles but identifying autistic behaviours based on their behavioural patterns. It is important to note that, the use of technology is not intended to replace the expert, instead we see a potential role of automated measures of movement to support clinicians by providing additional objective and accurate information, in a time-efficient way. Our early exploration indicates that this is a genuine possibility for future clinicians and autism practitioners.

# 5 Chapter Five: General Discussion

There were two main objectives in this thesis. The first objective was to examine the role of Emotion Regulation in the relationship between Executive Functioning and Restricted and Repetitive Behaviours (RRBs) and to test the hypothesis whether Emotion Regulation is the mediator in this relationship. The second goal was to develop an automated measurement and test its usability in measuring repetitive behaviours in autism. In this final chapter, the main findings of this thesis will be summarized before considering implications and future directions.

## 5.1 The role of Emotion Regulation in the relationship between Executive Functioning and RRBs

### 5.1.1 Why is the relationship between RRBs, Executive Functioning, and Emotion Regulation important?

Children with Autism Spectrum Conditions (ASC) and their families may experience challenges in their daily life resulting from RRBs, executive dysfunction and deficits in Emotion Regulation. Caregivers of children with ASC have rated RRBs as the most challenging characteristics in ASC (Bishop et al., 2007) which frequently result in higher stress levels for families (Lecavalier et al., 2006). These behaviours are said to interfere with a child's development of functional skills (Cuccaro et al., 2003), and create social stigma that may reduce learning opportunities for social interaction with peers (Nadig et al., 2010). Similarly, executive dysfunction and emotion dysregulation are common challenges experienced by individuals with ASC and have been reported to have negative impact on both individual's and family's functioning. For example, individuals with ASC have been observed having difficulty in switching from one activity to another (Demetriou et al., 2019) and difficulty in regulating their emotion using adaptive strategies (Costescu et al., 2016). We believe that further understanding the relationship between these constructs is necessary to further our theoretical understanding of the challenges related to them. However, these aspects received considerably less attention than other characteristics of ASC.

### 5.1.2 ASC in Malaysia

A "zero reject" policy was implemented in Malaysia in January 2019 to ensure all special needs children access education with 'reasonable accommodation' to meet their individual needs (Gan, 2019). To accommodate this large number of special needs children in inclusive education settings demands not only a strong underpinning implementation plan, but also educators who possess adequate knowledge of various disorders and disabilities. The education ministry has allocated RM54 million "to provide facilities for special needs children" and another RM16 million in the beginning of 2020 for 320 schools to improve the special needs classroom (MBK) infrastructure (Bernama, 2020). The awareness of autism has increased among Malaysians (Dolah et al., 2011) but studies have found Malaysians still have low exposure and actual knowledge regarding specific aspects of autism (Williams et al., 2011). In addition to fundings provided in education settings, research is essential for betterment of the autism society in a developing country like Malaysia but research into ASC is primarily carried out in Western nations.

Despite the fact that the core autism characteristics are believed to be universal and almost all the findings in this thesis might not seem to be culturally specific, there is preliminary evidence suggesting the influence of cultural differences over the identification and/or reporting of symptomatology (Freeth et al., 2013, 2014; Norbury & Sparks, 2013). For example, parents and caregivers in the Eastern communities (e.g., in the Japanese communities), do not interpret a lack of interest in other children as autism symptomatology as it does in the Western communities (e.g., in the USA communities) but a display of modesty or shyness (Inada et al., 2011). Assessment tools for autism that have primarily been developed in the Western nations (e.g., AQ, Baron-Cohen et al., 2001; CHEXI, Thorell & Nyberg, 2008); *RBQ-2,* Leekam et al., 2007) may affect the validity and reliability if used in Eastern communities. Freeth et al. (2013) examined the expression of autistic traits in a sample of typically developing young adults from one Western culture (UK) and two Eastern cultures (Malaysia and India) using the AQ and found Malaysian and Indian populations scoring higher. The researchers suggested that certain AQ items were interpreted differently in Eastern cultures resulting in differences how behaviours are perceived. Another study found four items in the AQ indicated potential cultural differences when compared the expression of autistic traits across India, Japan and the UK (Carruthers et al., 2018). There has also been a continuing oral discussion during autism conferences in Malaysia regarding the word choice and comprehension issues of the items in parent-report measures developed in the Western nations. Although these tools are increasing being used in other cultural contexts (Soto et al., 2015; Stewart & Lee, 2017), adaptions are indispensable when researchers are using them with non-Western populations to minimize the cross-cultural differences, especially when translation is being conducted.

### 5.1.3 What were the aims and what has this thesis achieved?

As mentioned previously, RRBs, Executive Dysfunction and Emotion Dysregulation are common challenges observed in individuals with ASC. The relationship between Executive Functioning and Emotion Regulation has been found significant in past studies (Carlson & Wang, 2007; Fuster et al., 2009; Li et al., 2020). Executive Functioning (Iversen & Lewis, 2021) and Emotion Regulation (Willemsen-Swinkels et al., 1998) have also been found significantly related to RRBs individually. Therefore, it is plausible to suggest a relationship between Executive Functioning, Emotion Regulation and RRBs in autism. However, at the time of writing this thesis, this two-way relationship has never been examined in ASC. The main goal of the first part of this thesis was to examine this relationship and the role of Executive Functioning and Emotion Regulation in RRBs.

In Chapter 2, we examined the relationship between RRBs, Executive Functioning and Emotion Regulation across 31 typically developing (TD) children and 33 ASC children. The first objective in this chapter was to examine the scores of these constructs across TD and ASC children with parent-report questionnaires. The results indicated that relative to TD children, children with ASC had higher levels of autistic traits and RRBs, as well as poorer Executive Functioning and Emotion Regulation skills. As hypothesized, these findings supported almost all the past studies which examined RRBs (Harrop et al., 2014; S. Leekam et al., 2007a), Executive Functioning (Demetriou et al., 2018; Hill, 2004; Lee et al., 2021), and Emotion Regulation (Mazefsky et al., 2013; Patel et al., 2017; Shaffer et al., 2022) in autism. The second objective of Chapter 2 was to examine the relationship between Executive Functioning and Emotion Regulation. As reported by parents, children with higher levels of Executive Functioning had better Emotion Regulation across both TD and ASC groups. As discussed throughout this thesis, significant relationships have been observed between Executive Functioning and RRBs, Emotion Regulation and RRBs, as well as Executive Functioning and Emotion Regulation separately. It is unclear whether the two-way relationship between these constructs is significant when put together. Given that Executive Functioning and Emotion Regulation are significantly related to RRBs independently, while significantly relating to each other, we hypothesized roles played by them in RRBs. These led to the main contribution of this chapter, providing a novel exploration into the roles of Executive Functioning and Emotion Regulation on RRBs using Hayes' mediation analysis model 4. Interestingly, we found Emotion Regulation was demonstrated as a mediator in this relationship in children with ASC but not TD children. These findings suggested that a decline in RRBs may be observed in ASC but not TD children when children demonstrate better Executive Functioning in which result in better Emotion Regulation. In another perspective, RRBs is related to the ability of regulating emotion, which may also serve as an alternative emotion regulation mechanism in the absence of adaptive emotion regulation strategies. The ability to regulate emotion can be improved by enhancing the levels of Executive Functioning.

It is worth mentioning that the findings from Chapter 2 were collected using parent-report questionnaires as the study was conducted during the Covid-19 pandemic when social restrictions were in place. Although parent-reported questionnaires have frequently provided meaningful findings in autism studies, we desired to gain further insight into the mediating role of Emotion Regulation in Executive Functioning and RRBs using observation and performance-based measures. Therefore, in Chapter 3, we manipulated the emotional contexts and observed RRBs across three conditions: Task, Neutral and Preferred. Participants were given three neuropsychological tasks during the Task condition. During a separate session, the participants were presented with preferred and neutral videos for 10 minutes in a counterbalanced order.

There were three objectives in Chapter 3. First, we explored the duration of RRBs across the three conditions. Children with ASC displayed significantly more repetitive behaviours in all three conditions compared to TD children. Children with ASC displayed most repetitive behaviours during the Preferred condition whereas TD children displayed most repetitive behaviours during the Neutral condition. These interesting findings reject the traditional proposition of RRBs as purposeless behaviour (Bodfish et al., 2000) and suggest that RRBs are likely to serve different functions in children with ASC and TD children. RRBs seem to serve as a regulatory mechanism during positive arousal for children with ASC and counter under-stimulation for TD children. As mentioned previously, some children with ASC walked away, looked away and engaged in self-injurious behaviours during the Task condition, which implies that children with ASC might engage in other behaviours, rather than RRBs, during negative arousals.

In addition to RRBs, the past studies (e.g., Lory et al., 2020; Machalicek et al., 2007) have also observed children with ASC engaged in behaviours such as elopement and self-injurious behaviours to escape from a difficult task. Over the past half-century, researchers have identified medical and behavioural factors as major contributors to challenging behaviours commonly associated with ASC (Edelson, 2022). The behavioural model proposed four predominant functions of behaviour: attention, escape, access, and automatic reinforcement (Cooper et al., 2019). Children with ASC, especially those with more severe social communication deficits (for example, those who cannot request for a break appropriately) often resort to behaviours to escape a difficult task (Matson et al., 2009). Based on the operant conditioning paradigm of challenging behaviours, the behaviours will be more likely to occur under similar circumstances if they are reinforced. For instance, individuals are more likely to engage in the challenging behaviours to escape a task if tasks have been removed from them in the past when they displayed the behaviours. On top of the behavioural model, the findings of this thesis highlight the importance to take the potential underlying mechanisms Executive Functioning and Emotion Regulation into considerations not only in RRBs research, but also in research that desire to understand challenging behaviours in ASC. The topographies of behaviour may vary even though they serve similar functions and have similar potential mediators.

In Chapter 3, we also replicated Chapter 2 and examined the relationship between the three constructs: Executive Functioning, Emotion Regulation and RRBs with rating scales. We found similar patterns as the previous findings in which Emotion Regulation serve as mediator in the relationship between Executive Functioning and RRBs in the total sample and the ASC sample. Then, we extended the study and replaced rating scales with neuropsychological tasks for Executive Functioning and observation for repetitive behaviours. The mediation path was no longer significant and there was no significant path from Emotion Regulation to RRBs in the total, TD and ASC groups. When all constructs were measured by rating scales, parents might have the tendency to complete the questionnaires having the impression that Emotion might be related to RRBs, which might be affected by Executive Functioning. On the other hand, neuropsychological tasks might be more sensitive to subtypes of Executive Functioning and therefore result in different patterns of findings. It is also important to note that, the duration of RRBs (in percentage) during the Neutral condition was used for analysis to minimize emotional valence but the post-hoc analysis found the RBQ-2 was significantly related to the duration percentage of RRBs during the Preferred condition only. However, the mediation analysis was carried out with the duration of RRBs (in percentage) during the Preferred condition, the patterns of finding were similar to the Neutral condition and the Task condition. This finding suggested that the emotional valence on RRBs in each condition may not as impactful as what we deduced.

The parent-report rating scale, CHEXI, was also not significantly correlated with the composite variable of neuropsychologist tasks of Executive Functioning, supporting some past studies that stated rating measures might assess different Executive Functioning constructs in relative to the neuropsychological measures (Mcauley et al., 2010; Thorell et al., 2010). This finding further explains why different patterns were found in the mediation analyses when different methods were used. Nevertheless, the rating measure, CHEXI, and the composite variable of neuropsychological task of Executive Functioning were significantly related with Emotion Regulation, implying that even if different constructs were examined using these measures, they are related to Emotion Regulation. Perhaps Emotion Regulation involves a variety of strategies and different Executive Functioning constructs might be related to different Emotion Regulation strategies. Past studies such as McRae et al.

(2012), Opitz et al. (2014), and Schmeichel et al., (2008) have also found Emotion Regulation strategies such as reappraisal and suppression related to different Executive Functioning constructs like working memory and cognitive flexibility in the typically developing (TD) population. For example, Schmeichel et al. (2008) found individuals with higher working memory capacity appraised emotional stimuli better. On the basis of the earlier and current findings, researchers may expect individuals who are better in a particular Executive Functioning construct such as working memory, to employ a specific emotion regulation strategy, like reappraisal, more capably than the other strategies (e.g., escape). Clinicians may also expect these individuals to benefit from cognitive-oriented treatment strategies, such as cognitive behavioural therapy which involves cognitive reappraisal that encourages individuals to stop their automatic negative thoughts and to develop alternative thoughts, more readily than those who are weaker in working memory.

As mentioned previously, Mazefsky et al. (2013) reported only 15 articles were produced following a PsychInfo search with the terms “emotion regulation” and “autism” in 2012, which then increased to 170 articles when we used the same search strategy in 2020. Although there is a growing number of studies, Emotion Regulation is still less studied in the autistic population than the TD population. At this point, the relationship between Executive Functioning constructs and Emotion Regulation in the autistic population is still unknown. This thesis found a similar pattern of relationship, as from the past studies of TD population, between Executive Functioning and Emotion Regulation in the ASC group not only when the parent-report rating scales (Chapter 2 and 3) were used but also with neuropsychological testing (Chapter 3). Further analyses on the relationship between Executive Functioning constructs and Emotion Regulation found that Emotion Regulation was related to the Working Memory and Inhibitory Control constructs from the CHEXI and was also related to the IES of DCCS and the IES of Go/No-Go in the total sample and ASC group. On the other hand, Emotion Regulation was only related to the Working Memory and Inhibitory Control constructs from the CHEXI in the TD group. As suggested previously, the results in the TD group must be interpreted cautiously as the task difficult level of the DCCS and Go/No-Go might not be appropriate. On the basis of the current results, it is plausible that specific Executive Functioning constructs might be related to specific Emotion Regulation strategy in ASC as in the TD population.

## 5.2 Automated Measurement for Repetitive Behaviours

### 5.2.1 Why is it important to have an automated measurement for repetitive behaviours?

Autism studies have increased rapidly over the past decade following the growing prevalence rates. An extensive range of measurements have been developed for measuring one of the hallmark characteristics of autism, Restricted and Repetitive Behaviours (RRBs), from paper-and-pencil rating scales to video-based coding. However, there is still a lack of standardised measurements, perhaps due to the broad variation of repetitive behaviours. Following Chapter 3, we recognised the need of having a time-effective, convenient and automated measurement for identifying, detecting and analysing repetitive behaviours in autism. As mentioned earlier in Chapter 4, the progression of technology has brought new measuring techniques which facilitate automated pose estimation and movement detection. Although there is still no research using this technology to analyse repetitive behaviours in autism, we believe this is a promising beginning for researchers in autism studies.

### 5.2.2 What were the aims and what has this thesis achieved?

In Chapter 4, a recently developed automated pose estimation technology, Open Pose, was modified so that angular data can be collected and analysed automatically using this tool, namely OpenPose_Angle. The first objective in Chapter 4 focused on comparing the estimation of keypoints between OpenPose_Angle and manual placement by participants from the Expert, Researcher and Non-Expert group. In this study, two arm positions were repeated for 5 times to generate repetitive movements having one arm moving from 90-degree elbow angle to 180-degree elbow angle. The results indicated that the angular data collected based on the keypoints generated by the OpenPose_Angle were more similar to the data collected from the keypoints that were manually placed by the Expert and Researcher groups but not the Non-Expert group. This is understandable as the participants from the Non-Expert group were not trained in research nor movement analysis. This finding has also suggested that the body keypoint placements generated by the OpenPose_Angle were reliable and consistent.

The second objective in Chapter 4 was to compare the angular data of typically developing individuals performing Jumping Jacks using OpenPose_Angle and Kinovea. Jumping Jacks were selected as the target movements because they are a set of movements that involves similar repetitive actions. These movements were performed by four different age groups: Child, Teenager, Young Adult and Elderly. Line graphs were generated using the data collected from the OpenPose_Angle and Kinovea and a Python-based algorithm was used to detect the peaks in the graph to calculate the frequency of the movement cycle. We found that the angular data collected from the OpenPose_Angle were relatively larger than the angles measured manually by the Kinovea. Apart from that, OpenPose_Angle provide more information in general, and consequently the automatic peak detection of the movement cycles could be easier for the OpenPose_Angle compared to Kinovea as the graph showed more apparent peaks. The results also showed that the sensitivity of the OpenPose_Angle was high across all groups and the specificity was high in almost all groups except the Child group. This is a sensible finding as pose estimation on children's movements have always been a huge challenge in Deep Learning due to limited datasets (Farinella et al., 2017). Children pose estimation can be important because it can enable new research and insights into child development and behaviours. By analysing the body postures and movements of children in different contexts and environments, researchers can better understand how children learn, interact with their surroundings, and develop skills and abilities. Accurate and real-time monitoring of children's behaviours may also help early identification of developmental delays so that children can access evidence-based interventions to mitigate their levels of disabilities.

Finally, the third objective in Chapter 4 was to utilize the OpenPose_Angle to measure the random repetitive motor movement displayed by children with ASC and these data were compared to the data gathered from the Kinovea. Despite OpenPose_Angle showing consistency and reliability on similar and repetitive movements performed by typically developing individuals, only 10% of data were usable from the spontaneous and random repetitive behaviours displayed by children with ASC. There were a few challenges in this study. As mentioned previously, the pose estimation on children's movements were more difficult than movements displayed by other age groups, not to mention prompt and random movements displayed by children with ASC. Apart from that, the OpenPose_Angle is dependent on detecting children's faces to form keypoints for other body parts and hence, the tool could not detect and analyse the movement whenever their face was away from the camera. During the experiment, children with ASC were more likely to look in different directions rather than the given tasks as they easily got distracted. When they were engaging in repetitive behaviours, they did not face the camera throughout. These reasons affected the data collection, and it is believed that having more cameras from all angles should provide better datasets.

## 5.3 Strengths and Limitations

Previous studies have investigated the individual relationship between Executive Functioning and Emotion Regulation (Carlson & Wang, 2007; Fuster et al., 2009; Li et al., 2020)., Executive Functioning and RRBs (Boyd et al., 2009; Perry et al., 2022; South et al., 2007), as well as Emotion Regulation and RRBs (Martínez-González et al., 2021). This thesis is the first to study these constructs altogether and explore the relationship between Executive Functioning, Emotion Regulation and RRBs. This relationship was not only tested with parent-report questionnaires but also neuropsychological and observational-based measures. Exploring the relationship between these constructs altogether can be important in furthering our theoretical understanding.

This thesis is also the first to modify a cutting-edge automated pose estimation technology for measuring repetitive behaviours in autism. The measurement for examining repetitive behaviours in autism often range from paper-and-pencil rating scales to video-based coding. There is still a lack of standardized measurement for these behaviours possibly due to multiple reasons such as a broad variant of repetitive behaviours, time-effectiveness, practicality and accessibility of the measurement. We believe that having an automated measuring tool that is time-effective and convenient facilitate the progression of science in autism research.

However, the work of this thesis was limited by a number of factors which have been referred to in each chapter. These factors constrain the conclusions and are reviewed here again. The first major limitation of this thesis was power issues. Mediation analysis tends to have lower power (Biesanz et al., 2010; Memon et al., 2018; Pan et al., 2018) in general because this analysis involves testing multiple hypotheses: the relationship between the independent variable and the mediators, the relationship between the mediators, the relationship between the mediators and the dependent variable, as well as the indirect effect of the independent variable on the dependent variable through the mediators. Additionally, mediation analysis involves more complex models and requires more assumptions than a simple regression analysis, which also contribute to lower power. In Chapter 2, the analysis found an acceptable power of .83 in the total sample size (*n* = 58) and .88 (*n* = 27) for the ASD group but extremely low power of .10 for the TD group (*n* = 31). The interpretation of the results for individual groups was constrained by the small sample size but sufficient for conducting exploratory factor analysis as with the total sample size of 58 participants. For the following study (as described in Chapter 3), we calculated sample sizes a priori based on the results collected from Chapter 2 and we recruited participants accordingly. The parent-report rating analysis found a power of .76 in the total sample (*n* = 114) and lower power of .36 was found in the total sample when neuropsychological tasks and observational measure were used. Mediation analysis can be sensitive to the quality of measurements (Liu & Wang, 2021; VanderWeele et al., 2012) and if there is a significant amount of measurement error or insensitivity in the mediator or the dependent variable, it can reduce the ability to detect significant indirect effects. In Chapter 3, we believe both measurement error and measurement insensitivity affect the power of the mediation analysis despite more participants were recruited in that study. Measurement error, specifically the inherent variability of the participants in the TD group, contributes to lower power in the study in Chapter 3. There were 19 participants (out of the 60 TD participants) who were attending remedial classes for additional academic support. Although their Executive Functioning and Emotion Regulation were not significantly different from the rest of the participants in the TD group, there were no significant differences between their Executive Functioning and Emotion Regulation and the ASC group. Additionally, another reason can be measurement insensitivity. Although inadequate sample size (as mentioned in Chapter 3) can contribute to measurement insensitivity, we also observed high degrees of within-group variability in both neuropsychological and observational measures. The IES of DCCS reported an extremely high degree of variance in the TD group, in relative to the parent-report measures. The total durations of RRBs (in percentage) and across Task, Preferred and Neutral conditions also have higher degree of variance than the RBQ-2 in both TD and ASC groups. Consequently, meaningful differences between groups can be difficult to detect due to the inherent variability issues and the high degrees of within-group variability, even if those differences can be theoretically meaningful.

The insensitivity of the OpenPose_Angle for random and spontaneous repetitive behaviours contributes to the second limitation in this thesis. Only 10% of complete data on random repetitive behaviours was successfully collected in Chapter 4. One of the reasons is that participants had to face the camera continuously throughout the session to ensure their movements being captured and analysed as the Open Pose algorithms use face landmark or detection localization. There are several ways to achieve this goal. For example, we can increase the number of cameras along with a camera synchronised unit that allow the participants' movements being captured from multiple directions and generate a more complete dataset for analysis but this method can be extremely expensive and impractical. Additionally, we can also modify the algorithms without relying on face landmark localization. At this point, however, only one study has proposed a novel real-time six degrees of freedom (6DoF) 3D face pose estimation technique that works without face detection or landmark localization (Albiero et al., 2020). The development of this technique is still at its beginning stage which has not been released publicly. Although the development of a novel technique that can address the limitation of the Open Pose may take a lot of research, effort, and time, we still believe it's pragmatically worthwhile given that this technology will not only advance scientific knowledge in autism research by enhancing data collection and increasing its efficiency, accuracy and precision, it also has the potential to revolutionize the way we understand RRBs and address the challenges faced by children with ASC and their families.

## 5.4 Implications and Future Directions

RRBs can be an important behavioural target in early intervention given that these behaviours might result in significant challenges such as dominating the individual's daily life (Harrop et al., 2016), interfering with the development of functional skills (Cuccaro et al., 2003) and social engagement (Loftin et al., 2008; Nadig et al., 2010). These behaviours also often lead to higher stress levels for parents and family members (Lecavalier et al., 2006) and affect the quality of family well-being (Lounds et al., 2007). Evidence-based interventions for lower order RRBs include Response Interruption and Redirection (RIRD; Ahearn et al., 2007; Liu-Gitz & Banda, 2009), Response Cost Procedures (Athens et al., 2008; Sidener et al., 2005), Environmental Enrichment Strategies (Piazza et al., 2000; Rapp, 2006, 2007), and Functional Communication Training (FCT; Kennedy et al., 2000). However, not one of the preliminary interventions involved teaching the individuals Executive Functioning and Emotion Regulation skills. Findings from the work of this thesis demonstrated possible mediating effect of Emotion Regulation in the relationship between Executive Functioning and RRBs suggest that teaching and improving Executive Functioning might improve one's ability to regulate emotion which might contribute to the management of RRBs. Li et al. (2020) developed a 2-month (12 sessions in total) Executive Functioning training curriculum based on four Executive Functioning sub-components: 1) inhibitory control, 2) cognitive flexibility, 3) working memory, and 4) problem solving, which has shown to improve emotional competence of typically developing preschool children. Given the relationship that was found in this thesis, we believe that providing both Executive Functioning and Emotion Regulation trainings are likely to provide individuals with ASC more adaptive skills to manage their RRBs.

In RIRD, individuals with ASC are physically or verbally blocked from engaging in RRBs; whereas in Response Cost Procedures, a positive consequence is removed if they engage in RRBs. These behaviour interventions have been deemed abusive and have been criticized by adults with ASC because they believed that individuals with ASC engaged in repetitive behaviours to calm themselves down or to cope with a stressful situation (Kapp et al., 2019). In stressful situations, stopping individuals from stimming can be considered as removing their ability from calming themselves down and adults with ASC believed that "people should be allowed to do what they like" (Kapp et al., 2019), although the participants also reckoned that some of the RRBs caused physical harm and might have negative influence on self and others. Not only that the work of Kapp et al.'s (2019) study further supports the findings of this thesis that RRBs might serve self-regulatory and emotion regulation purposes, it also supports the fact that we can simply improve individuals' Emotion Regulation skills so that they can develop an adaptive skill to regulate and calm themselves down without resulting in any negative consequences.

Observational measures used to collect data during the manipulation of emotional contexts found that children with ASC generally demonstrated more repetitive movements than TD children. TD children engaged in more repetitive movements during the Neutral condition whereas children with ASC engaged in more repetitive movements when they were presented with preferred items. Besides repetitive movements, children with ASC also demonstrated behaviours such as leaving seats, removing their attention from task and self-injurious behaviours during the Task condition. The fact that repetitive behaviours potentially serve different functions for TD and ASD children speaks to the issue of whether these behaviours are best construed as a collection of Emotion Regulation strategies or just adaptive strategies to regulate imbalanced state of being under-stimulated. The current findings lend more support to homeostatic regulations in general even for children with ASC as the duration of RRBs (in percentage) was also high in the Neutral condition. On the basis of these findings, we would predict that both TD children and children with ASC are more likely to engage in RRBs when they are under-stimulated and are likely to engage in other behaviours to regulate their emotion especially in Task condition.

The development of OpenPose_Angle is still at its beginning stage and is not ready for measuring random repetitive movements in less controlled environments. To develop a reliable and effective measurement tool that involve novel techniques requires a lot of time, effort and research. However, we believe the existence of an automated measurement tool for repetitive behaviours not only can improve the quality of life for individuals with autism but also provide objective data for clinician during assessment and intervention. This technology can be adapted and used in ecological settings for autism research and intervention in several ways. For example, OpenPose_Angle can be utilized to automatically track and analyse the body movements and postures of individuals with autism spectrum disorder (ASD) in naturalistic settings. Researchers can use this data to study patterns of movement, social interactions, and sensory-motor behaviours in real-world contexts, providing insights into the ecological validity of findings. OpenPose_Angle can also assist in the assessment and diagnosis of autism by quantifying specific behavioural characteristics and subsequently, clinicians and researchers can identify subtle motor abnormalities that may be indicative of autism. Last but not least, OpenPose_Angle can also support the development and evaluation of behavioural interventions for individuals with autism by providing objective feedback on behavioural patterns. Therapists and educators can use real-time feedback from OpenPose_Angle to promote the development of motor skills, social communication, and adaptive behaviours in ecological settings.

This measurement tool can be used to collect data to identify the behavioural patterns for different repetitive behaviours, automatically and support individuals' daily life.

## 5.5 Conclusions

This thesis has provided significant contributions to the literature on RRBs, Executive Functioning and Emotion Regulation in ASC, despite some of the limitations. There are several unique contributions of this thesis: 1. children with ASC engaged in more repetitive behaviours during the Preferred condition whereas TD children engaged in more repetitive behaviours during the Neutral condition, suggesting that repetitive behaviours potentially serve different functions for these two populations; 2. for the first time in the literature on RRBs, the two-way relationship between Executive Functioning, Emotion Regulation and RRBs was examined, and Emotion Regulation was considered as a possible mediator for RRBs in ASC; 3. the mediating effect of Emotion Regulation in ASC was significant when parent-report scales were employed but not neuropsychological tasks and observational measure; 4. the OpenPose_Angle was able to provide kinematic data of repetitive movements performed by typically developing individuals in a shorter time, with high sensitivity and specificity, in relative to Kinovea; 5. but it is still not ready for gathering data of random and spontaneous repetitive behaviours displayed by children with autism.

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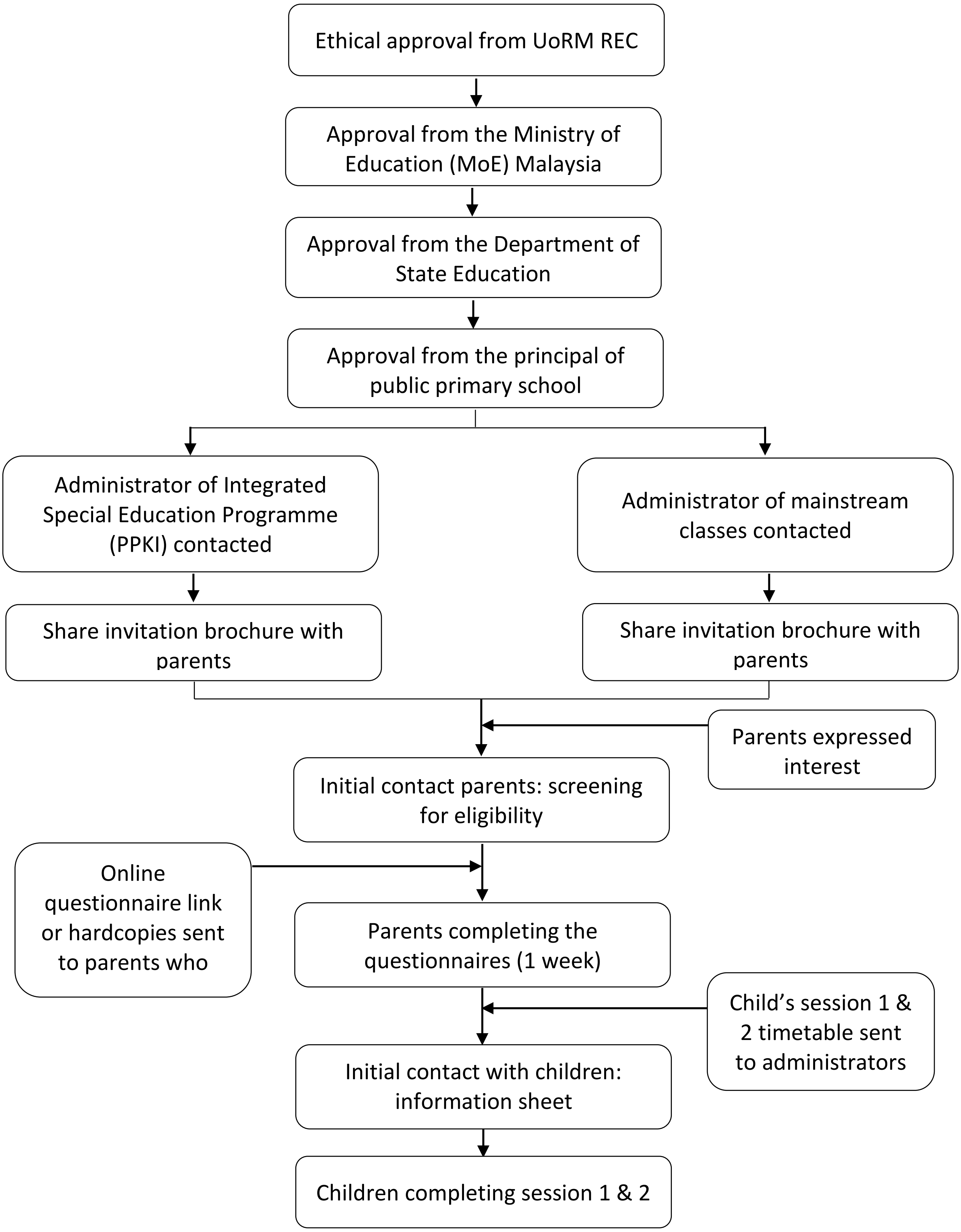
## Appendices

## Appendix 1

*Schematic representation of the recruitment process*



# Appendix 2

**INFORMATION SHEET**
**(Session 1)**

**INFORMATION SHEET**
**(Session 2)**

# Appendix 3

## Demographic Questionnaire

Demographic Information

1. Parent or caregiver's name: ________________
2. Child's name: ________________
3. Child's school and class: ________________
4. Child's date of birth (DD/MM/YYYY): ________________
5. Child's gender: ________________
6. Contact or WhatsApp number: ________________
7. Email: ________________

1. Has your child ever received a diagnosis of Autism Spectrum Disorder (ASD)?
    a. Yes
    b. No
    c. Prefer not to say

8. Has your child ever received a diagnosis of any other atypical development or learning problems?
    a. Yes
    b. No
    c. Prefer not to say
9. Referring to the previous question, if yes, what is the diagnosis?
10. Have any of your close family members (e.g., grandparents, parents, siblings, children, nieces, nephews, aunts, uncles, and first cousins) ever received a diagnosis of Autism Spectrum Disorder?
    a. Yes
    b. No
    c. May be

# Appendix 4

## Preference Assessment

You may answer the following questions together with your son, daughter, partner, or others who are also your child's caregivers, to provide the options that reflect your child's preference most appropriately.

1. List down 5 movies/cartoons that your child likes to watch.
2. List down 5 neutral movies or cartoons that your child neither likes nor dislikes strongly.
3. Which of the following activities does your child enjoy?

| | Strongly dislikes | Dislikes | Neutral | Likes | Strongly likes |
|---|---|---|---|---|---|
| Puzzles | ☐ | ☐ | ☐ | ☐ | ☐ |
| Bubbles | ☐ | ☐ | ☐ | ☐ | ☐ |
| Painting | ☐ | ☐ | ☐ | ☐ | ☐ |
| Balls | ☐ | ☐ | ☐ | ☐ | ☐ |
| Cars | ☐ | ☐ | ☐ | ☐ | ☐ |
| Playdough | ☐ | ☐ | ☐ | ☐ | ☐ |
| Animal figures | ☐ | ☐ | ☐ | ☐ | ☐ |
| Story books | ☐ | ☐ | ☐ | ☐ | ☐ |

4. Is there any other activities that you child strongly likes?

______________________________________________________________

# Appendix 5

Bahasa Malaysia (BM) Translation and English Back Translation for Waschbusch’s Parent-Report Cognitive Abilities, Pragmatic Abilities Questionnaire (PAQ) and Short Sensory Profile (SSP)

Waschbusch's Parent-Report Cognitive Abilities

| | | | |
|---|---|---|---|
| 1 | Has difficulty grasping how the parts should fit together in a puzzle | Mengalami kesukaran untuk memahami bagaimana kepingan teka-teki (puzzle) itu harus sesuai bersama | Have difficulty understanding how the pieces of the puzzle should fit together |
| 2 | Cannot tell time with dial watch | Tidak dapat memberitahu masa dengan jam tangan | Unable to tell the time |

| | | | |
|---|---|---|---|
| 3 | Does s/he have trouble describing or remembering the way things s/he has seen look when they are not present? | Mengalami kesukaran untuk menerangkan atau mengingati rupa sesuatu yang dilihatnya apabila tiada | Has difficulties in explaining or remembering an object they saw when it is not present |
| 4 | If given two facts, does s/he have trouble integrating into a new concept? | Jika diberi dua fakta, dia menghadapi masalah untuk memasukkan/mengintegrasikan ke dalam konsep baharu | If given two facts, they have trouble to incorporate/integrate new concepts |
| 5 | Cannot remember what s/he reads [is read to her/him for non-readers] | Tidak ingat apa yang dia baca [atau apa yang dibaca oleh pembaca lain kepadanya] | Does not remember what they have read [or been read to by someone else] |
| 6 | Has difficulty following the conversations of others | Mengalami kesukaran memahami perbualan orang lain | Has difficulty understanding other people’s conversation |
| 7 | Does s/he have difficulty following verbal instructions? | Adakah dia mengalami kesukaran mengikut arahan lisan? | Do they have difficulty understanding written instructions? |
| 8 | Does not seem to know how to talk to other children | Tidak tahu bagaimana untuk bercakap dengan kanak-kanak lain | Does not know how to speak to other children |
| 9 | Had trouble learning to cut with scissors | Menghadapi masalah belajar memotong dengan gunting | Have difficulties in learning how to cut with scissors |
| 10 | Has problems drawing a straight line | Menghadapi masalah melukis garisan lurus | Have difficulties drawing a straight line |
| 11 | Artwork looks like it was done by someone much younger | Karya seni kelihatan seperti dilakukan oleh seseorang yang lebih muda | Artwork looks like it has been done by someone younger |
| 12 | Seems very slow when using handwriting to copy material | Nampaknya sangat perlahan apabila menyalin kerja rumah | It seems very slow at copying homework |

| | | | |
|---|---|---|---|
| 13 | Seems to have trouble hearing the difference between similar sounding words | Tidak dapat membezakan antara perkataan yang sama bunyinya | Unable to distinguish between words with the same sounds |
| 14 | Seems to have trouble remembering which sounds go with which letters | Nampaknya sukar untuk mengingati bagaimana huruf sepadan dengan bunyinya | It seems difficult to remember how the letters correspond to their sounds |
| 15 | Seems to know fewer words than others his/her age | Nampaknya mengetahui lebih sedikit perkataan daripada orang lain seusianya | Seems to know fewer words than anyone else their age |
| 16 | Can read single words but gets confused when several words are together | Boleh membaca perkataan, tetapi menjadi keliru apabila beberapa perkataan disatukan | Able to read words, but gets confused when several words are combined |
| 17 | When asked to do more than one thing at a time, tends to forget what to do next | Apabila diminta melakukan lebih daripada satu perkara pada satu masa, sering terlupa apa yang perlu dilakukan seterusnya | When asked to do more than one task, they often forget what to do next |
| 18 | Has a difficult time remembering where things are | Mengalami kesukaran untuk mengingati di mana barangnya | Have difficulties in remembering where items are being kept |
| 19 | Remembers things for a short time, then forgets them | Mengingati sesuatu untuk masa yang singkat, kemudian melupakannya | Remembering something for a short time, then forgetting it |
| 20 | Has problems remembering the order in which events occurred | Mengalami kesukaran untuk mengingati urutan peristiwa | Have difficulties remembering the sequence of events |

| | | | |
|---|---|---|---|
| | | Pragmatic Abilities Questionnaire (PAQ) | |
| 1 | can make comments relevant to the topic of a conversation and complete it | boleh membuat dan melengkapkan ulasan yang berkaitan dengan topik perbualan | Able to make and complete comments relevant to the topic of conversation |
| 2 | is able to initiate conversation | boleh memulakan perbualan | Able to start a conversation |
| 3 | asks for clarification if s(he) does not understand something that is said to him/her | meminta penjelasan jika dia tidak memahami sesuatu yang dikatakan kepadanya | Ask for an explanation if they do not understand what is being said to them |
| 4 | uses eye contact while talking and/or listening. | menggunakan hubungan mata semasa bercakap dan/atau mendengar. | Able to maintain eye-contact when they are talking or listening |

| | | | |
|---|---|---|---|
| 5 | talks properly in routine daily conversations (for example: “Hello”, “How are you?”, “I’m fine, thanks”). | bercakap dengan betul dalam perbualan harian (cth: "Hello", "Apa khabar?", "Saya tidak apa-apa, terima kasih"). | Speaks correctly in everyday conversation (For example: “Hello”, “How are you?”, “I am fine, thank you”). |
| 6 | can make promises to others (for example “I promise to sleep at night”). | boleh membuat janji kepada orang lain (cth. "Saya berjanji untuk tidur pada waktu malam") | Able to make promises with someone (For example: “I promise to sleep at night”) |
| 7 | uses the politeness markers “please”, “thank you” and “excuse me” properly. | menggunakan tanda sopan “tolong”, “terima kasih” dan “maaf” dengan betul. | Uses polite signs “Please”, “Thank you” and “Sorry” correctly |
| 8 | requests more information if not understanding the topic. | meminta maklumat lanjut jika tidak memahami topik. | Asking for more information if they do not understand |
| 9 | uses facial expression, gestures or body movements to convey his/her feelings or thoughts. | menggunakan mimik muka, gerak isyarat atau pergerakan badan untuk menyampaikan perasaan atau fikirannya. | Use facial expressions, gestures or body movements to convey feelings or thoughts |
| 10 | is able to respond to questions. | mampu menjawab soalan. | Able to answer a question |
| 11 | introduces new topics in the discourse. | memperkenalkan topik baru dalam wacana. | Able to introduce new topics in speech |
| 12 | plays alone. | bermain sendirian. | Plays alone |
| 13 | seems to be perceived as odd and unusual by other people. | seolah-olah dianggap ganjil dan luar biasa oleh orang lain. | Seems to be considered odd an unusual by others |
| 14 | can agree or disagree with a topic of conversation. | boleh bersetuju atau tidak bersetuju dengan sesuatu topik perbualan. | Able to agree and disagree with a topic of conversation |
| 15 | asks questions when not knowing something. | bertanyakan soalan apabila tidak | Ask questions when they don’t know |

| | | | |
|---|---|---|---|
| | | mengetahui sesuatu perkara. | something |
| 16 | talks clearly about something that the listener does not know about. | bercakap dengan jelas tentang sesuatu yang tidak diketahui oleh pendengar. | Speak clearly about a topic that the listener does not know |
| 17 | can warn about something; for example, “take care that the door won’t hit your head”. | boleh memberi amaran tentang sesuatu; contohnya, "Berhati-hatilah pintu akan memukul kepala anda". | Able to give warning; For example: “Be careful, the door is about to hit your head” |
| 18 | follows the rules of games. | mematuhi peraturan permainan. | Able to follow rules of the game |
| 19 | repeats or explains it so that more information is conveyed to the listener when his/her intention has not been understood. | mengulangi atau menjelaskannya supaya lebih banyak maklumat disampaikan kepada pendengar apabila niatnya belum difahami. | Repeat or explain it so more information is conveyed to the listener when his intentions have not been understood |
| 20 | can produce long and complicated sentences. | boleh menghasilkan ayat yang panjang dan kompleks. | Able to create long and complex sentences |
| 21 | does not infer correct meaning from a speaker’s message and gives unusual responses as a result. | tidak menyimpulkan maksud yang betul daripada mesej penutur dan memberikan respons yang luar biasa akibatnya. | Did not deduce the correct meaning from the speaker and gives the speaker a usual response because of it |
| 22 | can understand sarcasm. | boleh memahami sindiran. | Able to understand sarcasm |
| 23 | The children let him/her to take part in group activities. | Kanak-kanak lain membenarkan dia mengambil bahagian dalam aktiviti kumpulan. | Other children allow him to participate in group activities |
| 24 | gives up the top of conversations. | melepaskan bahagian atas perbualan. | Let’s part of the conversation go |
| 25 | ends conversations in a correct manner. | menamatkan perbualan dengan cara yang | Ending the conversation correctly |

| | | | |
|---|---|---|---|
| | | betul. | |
| 26 | avoids talking to adults. | mengelak bercakap dengan orang dewasa. | Avoid talking to adults |
| 27 | understands indirect requests (for example, in response to the question “Would you like to eat with your hands?”, doesn’t answer “yes” or “no”, but washes his/her hands). | emahami permintaan tidak langsung (contohnya, sebagai jawapan kepada soalan "Adakah anda mahu makan dengan tangan anda?", tidak menjawab "ya" atau "tidak", tetapi mencuci tangannya). | Understand indirect requests (e.g.; in response to a question “Do you want to eat with your hands?”, They don’t answer “yes” or “no” but washes their hands) |
| 28 | understands other people’s emotions (for example, sadness, happiness and anger). | memahami emosi orang lain (contohnya, kesedihan, kegembiraan dan kemarahan). | Understand other people’s emotions (e.g.; Sadness, Happiness, and Anger) |
| 29 | can tell a story or describe what he/she has done in an orderly sequence of events. | boleh bercerita atau menerangkan apa yang telah dilakukannya dalam urutan peristiwa yang teratur. | Able to tell or explain what they have done in an orderly sequence |
| 30 | can talk differently in harmony with the context or needs of the listener (for example, talking differently to a child vs an adult). | boleh bercakap secara berbeza selaras dengan konteks atau keperluan pendengar (contohnya, bercakap berbeza dengan kanak-kanak vs orang dewasa). | Able to speak differently in accordance with the context or needs of the listener (e.g; speak differently with children vs. adults) |
| 31 | talks to other children when s(he) is with them. | bercakap dengan kanak-kanak lain apabila dia bersama mereka. | Talk to other children when they are with them |
| 32 | tries to negotiate with other people if they disagree with his/her ideas. | cuba berunding dengan orang lain jika mereka tidak bersetuju dengan ideanya. | Try to negotiate with others if they do not agree with their idea. |
| 33 | can understand idioms; for example, “wipe that smile off your face”. | boleh memahami simpulan bahasa; contohnya, "hapuskan senyuman itu dari wajah anda". | Able to understand idioms (e.g; Wipe that smile of your face) |

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|---|---|---|---|
| 34 | uses verbal behaviours, such as “yeah” and “really”, and non-verbal behaviours, such as head nods, smiling and looking, to give feedback to the speaker. | menggunakan tingkah laku lisan, seperti "ya" dan "sungguh", dan tingkah laku bukan lisan, seperti anggukkan kepala, tersenyum dan melihat, untuk memberi maklum balas kepada penceramah. | use verbal behaviours, such as “yes” and “really”, and non -verbal behaviors, such as nodding, smiling and looking, to respond to the speaker. |
| 35 | talks about his/her wishes in the future | bercakap tentang keinginannya pada masa hadapan | Talking about their future desires |
| 36 | seems inattentive, distant or preoccupied in the presence of familiar adults | Nampaknya tidak memberi perhatian, dingin, atau sibuk dengan kehadiran orang dewasa yang akrab | Seem to be inattentive, cold, or preoccupied with the presence of a familiar adult. |
| 37 | talks in a way appropriate for different characters when playing | bercakap dengan cara yang sesuai untuk watak yang berbeza semasa bermain | Speak in an appropriate manner for different characters while playing |
| 38 | is able to defend himself/herself by talking (for example, “this pencil is mine; give it back to me”) | mampu mempertahankan dirinya dengan bercakap (contohnya, “pensel ini milik saya, kembalikan kepada saya”) | Able to stand up for themselves (For Example: That pencil belongs to me, please give it back to me) |
| 39 | can understand what is not explicitly stated when s(he) listens to a narration (For example, “Ali’s father had already said to Ali “If you get a good score, I will buy a bike for you”. Ali is riding a bike now. Does he/she understand that Ali has scored?) | boleh memahami perkara yang tidak dinyatakan secara eksplisit apabila dia mendengar cerita (Contohnya, "Ayah Ali telah pun berkata kepada Ali "Jika kamu mendapat markah yang baik, saya akan membelikan sebuah basikal untuk kamu". Ali sedang menunggang basikal. Adakah dia faham bahawa Ali telah mencapai matlamatnya?) | Able to explicitly understand a detail in a story that was not made obvious when they are listening to a story (For Example: Ali’s Father spoke to Ali “If you get good grades, I will buy a bicycle for you”. Ali is riding a bicycle. Are they able to pick up that Ali achieved his goal?) |
| 40 | talks about his/her emotions | bercakap tentang emosinya | Talk about emotion |

## Appendix 6

Chinese Translation and English Back Translation for Waschbusch's Parent-Report Cognitive Abilities, Pragmatic Abilities Questionnaire (PAQ) and Short Sensory Profile (SSP)

| No | English Original | Chinese Translation (by Ling Chih Chong) | English Back Translation **(**by Yi Shan Wong) |
|---|---|---|---|
| | | Waschbusch's Parent-Report Cognitive Abilities | |
| 1 | Has difficulty grasping how the parts should fit together in a puzzle | 难以了解拼图中的各个部分应如何组合在一起 | Has difficulty understanding how the pieces of the puzzle fit together |
| 2 | Cannot tell time with dial watch | 不能依据表盘手表告知时间 | Cannot tell the time based on the clock and watch |
| 3 | Does s/he have trouble describing or remembering the way things s/he has seen look when they are not present? | 他/她是否难以描述或记住他/她看到的事物就算那个事物已经不在视觉范围内？ | Does he/she have difficulty describing or remembering what he/she sees even when it is out of sight? |
| 4 | If given two facts, does s/he have trouble integrating into a new concept? | 如果给予两个事实，他/她是否难以融入新概念？ | If given two facts, is it difficult for him/her to incorporate new concept? |
| 5 | Cannot remember what s/he reads [is read to her/him for non-readers] | 不记得她/他读了什么[或不记得其他读者读给她/他的] | Does not remember what she/he read (or what other readers read to her/his) |

| | | | |
|---|---|---|---|
| 6 | Has difficulty following the conversations of others | 难以听懂别人的对话 | Has difficulty understanding other's conversations |
| 7 | Does s/he have difficulty following verbal instructions? | 他/她是否难以听从口头指示? | Does he/she have difficulty following verbal instructions? |
| 8 | Does not seem to know how to talk to other children | 不知道如何与其他孩子交谈 | Does not know how to talk to other children |
| 9 | Had trouble learning to cut with scissors | 有困难学习用剪刀剪东西 | Has difficulty learning to cut things with scissors |
| 10 | Has problems drawing a straight line | 有困难画直线 | Has difficulty drawing straight lines |
| 11 | Artwork looks like it was done by someone much younger | 美术作品看起来像是由年纪更小的人完成的 | Artwork looks like it was done by someone younger |
| 12 | Seems very slow when using handwriting to copy material | 抄写作业时似乎很慢 | Seems to be slow when doing homework |
| 13 | Seems to have trouble hearing the difference between similar sounding words | 似乎 听不出发音相似的词 之间的区别 | Cannot seem to tell the difference between words that sound alike |
| 14 | Seems to have trouble remembering which sounds go with which letters | 似乎很难记住字母与其发音的搭配 | Seems to have difficulty remembering letters and their pronunciations |
| 15 | Seems to know fewer words than others his/her age | 似乎比他/她同龄的人知道的单词少 | Seems to know fewer words than people of his/her age |
| 16 | Can read single words but gets confused when several words are together | 能读单词，但当几个单词放在一起时会感到困惑 | Can read words, but gets confused when several words are put together |

| | | | |
|---|---|---|---|
| 17 | When asked to do more than one thing at a time, tends to forget what to do next | 当被要求一次做多过一件事时，往往会忘记下一步该做什么 | Often forgets what to do next when asked to do more than one thing at a time |
| 18 | Has a difficult time remembering where things are | 很难记住东西在哪里 | Has difficulty remembering where things are |
| 19 | Remembers things for a short time, then forgets them | 事情只记得一会儿，然后就忘记它们 | Only can remember things for a while, then forget them |
| 20 | Has problems remembering the order in which events occurred | 难以记住事件发生的顺序 | Has difficulty remembering the sequence of events |

| | | | |
|---|---|---|---|
| Pragmatic Abilities Questionnaire (PAQ) | | | |
| 1 | can make comments relevant to the topic of a conversation and complete it | 可以发表并完成与对话主题相关的评论 | Can express and complete comments related to the topic of the conversation |
| 2 | is able to initiate conversation | 能够开始对话 | Can start a conversation |

| | | | |
|---|---|---|---|
| 3 | asks for clarification if s(he) does not understand something that is said to him/her | 如果 他/她 不明白别人对 他/她 说的话，他/她会要求澄清 | If he/she does not understand what is being said to him/her, he/she will ask for clarification |
| 4 | uses eye contact while talking and/or listening. | 在说话和/或聆听时使用眼神交流。 | Uses eye contact when talking and/or listening |
| 5 | talks properly in routine daily conversations (for example: “Hello”, “How are you?”, “I’m fine, thanks”). | 在日常的日常对话中会好好说话（例如：“你好”、“你好吗？”、“我很好，谢谢”）。 | Speaks well in everyday conversations (e.g., "Hello", "How are you?", "I'm fine, thank you") |
| 6 | can make promises to others (for example “I promise to sleep at night”). | 可以向别人承诺（例如“我保证晚上睡觉”） | Can make promises to others (e.g., "I promise to sleep at night") |
| 7 | uses the politeness markers “please”, “thank you” and “excuse me” properly. | 正确使用礼貌标记“请”、“谢谢”和“对不起”。 | Make proper use of the polite signs "please," "thank you," and "sorry." |
| 8 | requests more information if not understanding the topic. | 如果不理解主题，会要求更多信息。 | Asks for more information if the topic is not understood |
| 9 | uses facial expression, gestures or body movements to convey his/her feelings or thoughts. | 使用面部表情、手势或肢体动作来表达他/她的感受或想法。 | Uses facial expressions, gestures or body movements to express his/her feelings or thoughts. |
| 10 | is able to respond to questions. | 能够回答问题。 | Can answer questions |
| 11 | introduces new topics in the discourse. | 引入或介绍新话题。 | Incorporates or introduces new topics |
| 12 | plays alone. | 独自玩 | Plays alone |
| 13 | seems to be perceived as odd and unusual by | 似乎被其他人认为是不一样或奇怪的。 | Seems to be perceived by others as |

| | | | |
|---|---|---|---|
| | other people. | | different or strange. |
| 14 | can agree or disagree with a topic of conversation. | 可以同意或不同意谈话的话题。 | Can agree or disagree with the topic of the conversation |
| 15 | asks questions when not knowing something. | 不知道的时候问问题。 | Asks questions when don’t know。 |
| 16 | talks clearly about something that the listener does not know about. | 听众不知道的事情会清楚地解释。 | Things the audience doesn't know will be explained clearly |
| 17 | can warn about something; for example, “take care that the door won’t hit your head”. | 可以警告某事；例如，“小心门会撞到你的头”。 | Can warn about something; for example, "Be careful that the door will hit your head" |
| 18 | follows the rules of games. | 遵守游戏规则。 | Complies with the rules of the game |
| 19 | repeats or explains it so that more information is conveyed to the listener when his/her intention has not been understood. | 在他/她（您的小孩）意图未被理解时，向听者传达更多信息，以便重复或解释它的意思 | When his/her (your child's) intent is not understood, convey more information to the listener in order to repeat or explain its meaning. |
| 20 | can produce long and complicated sentences. | 能制造长而复杂的句子。 | Can create long and complex sentences。 |
| 21 | does not infer correct meaning from a speaker’s message and gives unusual responses as a result. | 不能从说话者的信息中推断出正确的意思，并因此做出不寻常的反应。 | Unable to infer the correct meaning from the speaker's message and therefore react in an unusual way. |
| 22 | can understand sarcasm. | 能理解讽刺。 | Can understand sarcasm. |
| 23 | The children let him/her to take part in group activities. | 其他孩子们让他/她参加团体活动。 | The other children let him/her participate in group activities. |

| | | | |
|---|---|---|---|
| 24 | gives up the top of conversations. | 放弃话题的主导权。 | Gives up on dominating the conversation |
| 25 | ends conversations in a correct manner. | 以正确的方式结束对话。 | Ends the conversation the right way。 |
| 26 | avoids talking to adults. | 避免与成年人交谈。 | Avoids talking to adults。 |
| 27 | understands indirect requests (for example, in response to the question “Would you like to eat with your hands?”, doesn’t answer “yes” or “no”, but washes his/her hands). | 理解间接的请求（例如，在回答“你想用手吃饭吗？”的问题时，不回答“是”或“否”，而是洗手）。 | Understands indirect requests (e.g., washing hands instead of answering "yes" or "no" to the question "Would you like to eat with your hands?"). |
| 28 | understands other people’s emotions (for example, sadness, happiness and anger). | 了解他人的情绪（例如，悲伤、快乐和愤怒） | Understands one’s emotions (e.g., sadness, happiness, and anger). |
| 29 | can tell a story or describe what he/she has done in an orderly sequence of events. | 能有条不紊地讲述一个故事或描述他/她所做的事情。 | Can tell a story or describe what he/she does in an orderly way. |
| 30 | can talk differently in harmony with the context or needs of the listener (for example, talking differently to a child vs an adult). | 可以根据听众的语境或需求进行不同的对话（例如，对儿童和成人的不同的交谈）。 | Can have different conversations based on the context or needs of the audience (e.g., different conversations with children and adults). |
| 31 | talks to other children when s(he) is with them. | 当他/她和其他孩子在一起时，与他们交谈。 | Talks to other children when he/she is with them |
| 32 | tries to negotiate with other people if they disagree with his/her ideas. | 如果其他人不同意他/她的想法，他/她会尝试与他们谈判。 | If others disagree with his/her ideas, he/she will try to negotiate with them. |
| 33 | can understand idioms; for example, “wipe that smile off your face”. | 能听懂成语；例如，“不再洋洋得意”。 | Can understand idioms, e.g., "don’t be walking/floating on air" |

| | | | |
|---|---|---|---|
| 34 | uses verbal behaviours, such as “yeah” and “really”, and non-verbal behaviours, such as head nods, smiling and looking, to give feedback to the speaker. | 使用语言行为，如“是”和“真的”，以及非语言行为，如点头、微笑和看向说话者。 | Use of verbal behaviours such as “yes” and “really”, and non-verbal behaviours such as nodding, smiling, and looking at the speaker. |
| 35 | talks about his/her wishes in the future | 谈论他/她未来的愿望 | Talk about his/her future wishes. |
| 36 | seems inattentive, distant or preoccupied in the presence of familiar adults | 在熟悉的成年人面前显得注意力不集中、疏远或全神贯注于其他事物 | Being inattentive, distant, or preoccupied with other things in front of familiar adults. |
| 37 | talks in a way appropriate for different characters when playing | 演奏时以适合不同角色的方式说话 | Speak in a way that suits different characters while playing |
| 38 | is able to defend himself/herself by talking (for example, “this pencil is mine; give it back to me”) | 能够通过言语为自己辩护（例如，“这支铅笔是我的，把它还给我”） | Able to speak in defense of themselves (e.g., "This pencil is mine, give it back to me") |
| 39 | can understand what is not explicitly stated when s(he) listens to a narration (For example, “Ali’s father had already said to Ali “If you get a good score, I will buy a bike for you”. Ali is riding a bike now. Does he/she understand that Ali has scored?) | 能听懂听旁白时没有明确表达的内容（例如，“阿里的父亲已经对阿里说过“如果你考得好，我给你买辆自行车”。阿里在骑自行车时，他/她能够了解阿里已经达到目标了吗？） | Able to understand the content that is not clearly expressed when listening to the narration (e.g., "Ali's father has said to Ali, "If you do well in the test, I will buy you a bicycle". When Ali is riding a bicycle, can he/she understand that Ali has achieved the goal?) |
| 40 | talks about his/her emotions | 谈论他/她的情绪 | Talk about his/her emotions |

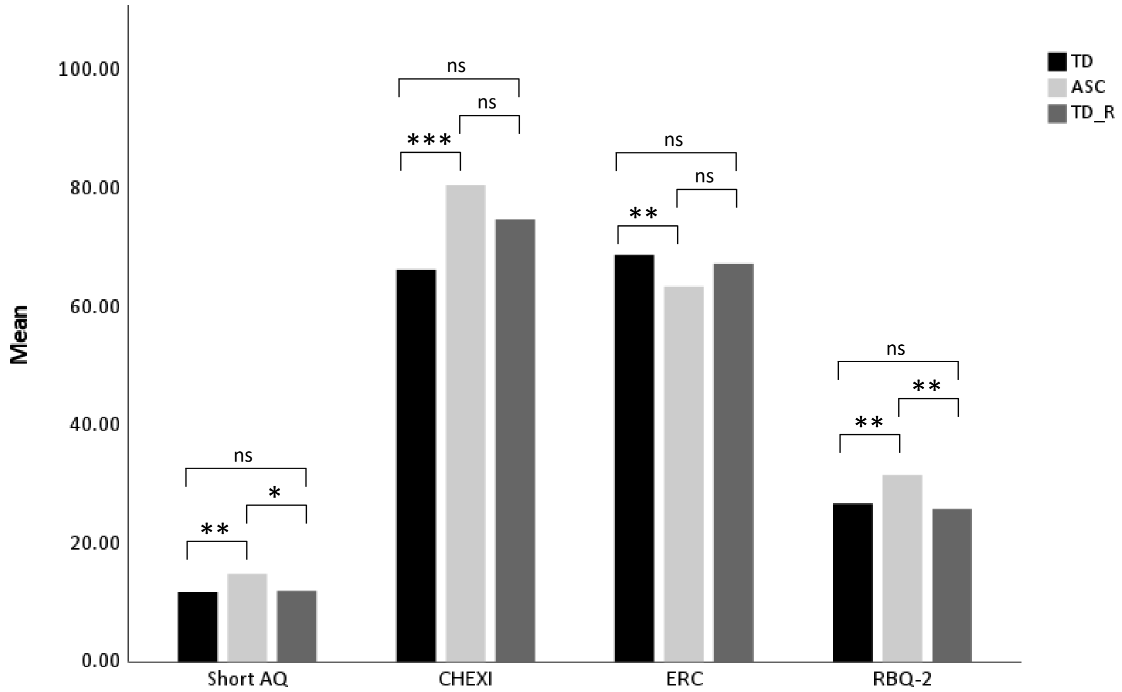
# Appendix 7

*Summary of Missing Data for Study 3*

| Questionnaire | <6% | 6-10% | 11-20% | 21-30% | 31-40% | >40% |
|---|---|---|---|---|---|---|
| AQ-Short | 8 | | | | 2 | |
| CHEXI | 1 | 2 | 1 | | | |
| ERC | 5 | 1 | | | | |
| RBQ-2 | 3 | | | | | |
| Waschbusch’s Scale | 2 | | | | | |
| SSP-2 | 2 | 1 | | | | |
| PAQ | 7 | 1 | 2 | 1 | | 1 |
| Go/No-Go | | | | | | 5 |
| DCCS | | | | | | 5 |
| Raven’s CPM | | | | | | 7 |

## Appendix 8

*Mean comparison of Parent-Report Measures (Short AQ, CHEXI, ERC, and RBQ-2) between TD, ASC and TD_R groups.*




*Note.* * $p < .05$, ** $p < .01$, *** $p < .001$

# Appendix 9

*Modified Python Code for OpenPose System*

| File Name | Domain | Function | Code |
|---|---|---|---|
| TfPoseEstimator | Neck | Distance from Center | dist_y = centers[pair[1]][1] - centers[pair[0]][1]<br>dist_x = centers[pair[1]][0] - centers[pair[0]][0]<br>The initial number 1 represents neck location, while 0 represents the origin.<br>The later number 1 represent y value, whereas 0 represents x value. |
| | | Angle (in degree) | neck_value = math.atan2(dist_y, dist_x) * 180 / 3.142 |
| | | Text | print ("Neck Value = ", neck_value) |
| | | Text Font | font = cv2.FONT_HERSHEY_SIMPLEX |
| | Right Shoulder | Angle (in degree) | rshoulder_value = math.atan2(dist_y, dist_x) * 180 / 3.142 - neck_value |
| | | Text on Image | cv2.putText(npimg, str(rad), centers[pair[0]], font, 0.5, common.CocoColors[pair_order], 1, cv2.LINE_AA) |
| | | Elipse | cv2.ellipse(npimg,(centers[pair[0]]), (20,20), neck_value, 0, rad, common.CocoColors[pair_order], 3) |
| | Right Elbow | Angle (in degree) | relbow_value = math.atan2(dist_y, dist_x) * 180 / 3.142 - rshoulder_value +180 |
| | | Text on Image | cv2.putText(npimg, str(rad), centers[pair[0]], font, 0.5, common.CocoColors[pair_order], 1, cv2.LINE_AA) |
| | | Elipse | cv2.ellipse(npimg, (centers[pair[0]]), (20, 20), rad+ rshoulder_value +180, 0, -rad, common.CocoColors[pair_order], 3) |
| | Left Shoulder | Angle (in degree) | lshoulder_value = math.atan2(dist_y, dist_x) * 180/3.142 - neck_value |
| | | Text on Image | cv2.putText(npimg, str (rad), centers [pair[0]], font, 0.5, common.CocoColors[pair_order], 1, cv2. LINE_AA) |
| | | Elipse | cv2.ellipse(npimg, (centers [pair [0]]), (20,20), neck_value +180, 0, rad, common.CocoColors [pair_order], 3) |

| | | |
|---|---|---|
| Left Elbow | Angle (in degree) | lelbow = math.atan2(dist_y, dist_x) * 180/3.142 - lshoulder_value - 180 |
| | Text on Image | cv2.putText(npimg, str (rad), centers [pair[0]], font, 0.5, common.CocoColors[pair_order],1, cv2.LINE_AA) |
| | Elipse | cv2.ellipse(npimg, (centers[pair[0]]), (20,20), lshoulder_value, 0, rad, common.CocoColors[pair_order], 3) |

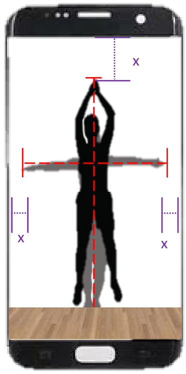
## Appendix 10

**Information sheet for video recording**

**1** Choose white or plain wall as background

**2** Place your phone on a tripod. The distance between your phone and your child can be varied as long as the phone's camera captures your child's whole body when (s)he is jumping.

Note: leave some spaces between the edge of phone and your child's body (see purple lines "x")




**3** Set your timer to 30 seconds.
Ask your child to start performing jumping jacks.
Start recording and start the timer.

**4** Stop recording after 30 seconds or when the timer rings.

**5** Check the video. Make sure you can see the whole body

**6** Repeat Steps 1-5 x 2.
You should have **THREE** 30-second videos of your child doing jumping jacks at the end of the session.

# Appendix 11

**Information sheet for children**

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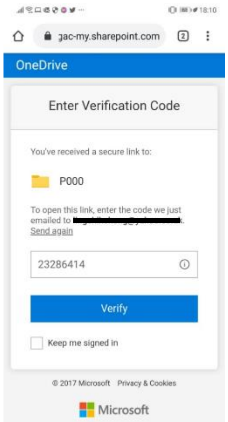
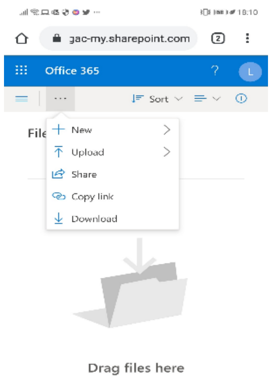
## Appendix 12

### Instructions for uploading videos

**1** You will receive a secure link from the researcher (e.g., https://livereadingac-my......). Click on the link and it will lead you a page where you have to enter a verification code that is sent to your email. Enter the code.




**2** After you enter the code, it will lead you into the One-Drive folder. Click on the three dots on your top left. Click "Upload".




**3** Click "Files".

**4** Click "Camera" or "Files". Choose videos that you have recorded or forms that you have signed. The title of the video/file will be shown in the folder when the upload is successful.