The Relationship between Theory of Mind and Traits

Associated with Autism Spectrum Condition and

Pathological Demand Avoidance Presentations

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Thesis declaration form

I confirm that the work presented in this thesis is my own. Where information has

been derived from other sources, I confirm that this has been indicated in the thesis.

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Overview

This thesis explores the relationship between theory of mind (ToM) and behaviour in childhood. It is presented in three parts.

Part 1 is a systematic literature review examining the relationship between ToM and aggressive behaviour in childhood. The review focuses on studies of typically developing children which report correlation analyses between ToM and aggression. Using a meta-analysis, the review found a weak but significant relationship between better-developed ToM and lower levels of aggressive behaviour.

Part 2, the empirical research paper, explores the profile of pathological demand avoidance (PDA) in autism spectrum conditions (ASC), and investigates the relationship between ToM and behaviours and traits associated with (1) ASC and (2) PDA. Quantitative data were collected via parent-report questionnaires and continuous and between-group analyses were conducted. Better parent-reported ToM was associated with lower levels of ASC traits, although no association was found between ToM and PDA traits. In addition, the findings support previous research arguing for the use of the PDA label to describe a set of symptoms within the autism spectrum. The results are discussed with reference to the wider literature and methodological limitations. This was part of a joint project with Anna Goodson, trainee clinical psychologist.

Part 3 is a critical appraisal on the research as a whole. The methodological challenges and limitations of the study are discussed. Broader conceptual issues are considered before concluding with further reflections on my own personal experience of the research process.

Impact Statement

The overall aim of this thesis was to contribute to an understanding of the relationship between theory of mind (ToM), and traits and behaviour in children with and without autism spectrum condition (ASC). ASC effects approximately 600,000 people in the UK. Higher rates of diagnosis in recent years have led to increased demands on services and the need for enhanced provision. There is significant heterogeneity in presentations of ASC, but related behavioural difficulties can significantly impact quality of life for many. ToM is a complex aspect of socio-cognition thought to be impaired in children with ASC, and associated with behavioural difficulties in both typically developing children and those with ASC. Therefore, an improved understanding of this relationship could help facilitate well-informed behavioural interventions for children with and without ASC, inform service delivery and provide further insight into traits and behavioural difficulties associated with ASC.

The literature review found that better ToM capacity relates to lower levels of aggressive behaviour in childhood, especially in children under the age of six. This finding helps to understand the underlying processes behind aggressive behaviour. In addition, it provides insight to inform the development of clinical interventions to reduce aggression in childhood, crucial to combatting the detrimental impact of inter-generational cycles of aggressive behaviour at an individual and societal level. The review also highlights important limitations with the sole use of informant-report measurement tools to assess behaviour, drawing attention to the need for further research in this area.

The empirical study found lower parent-reported ToM in children with ASC compared to controls, and that better ToM significantly predicted lower rates of social

and communication impairments in ASC. This provides evidence to suggest further investigation of interventions based on ToM training may be worthwhile in order support the development of social and communication skills in children with ASC – key to facilitating independence and contributing to an improved quality of life.

The empirical paper also studied pathological demand avoidance (PDA) traits in ASC. PDA traits in ASC are not well understood, but have been associated with increased rates of behavioural difficulty, parental stress and educational exclusion. There was no relationship between these traits and ToM. This suggests the mechanisms underlying these behaviours may differ to those implicated with core traits in ASC and stresses the need for professionals to develop distinct behavioural management and support strategies to manage these. In addition, the paper reveals much-needed insight into the behavioural profile of PDA, emphasising the importance of individualised assessment and early identification of traits during the ASC diagnostic process.

In conclusion, this thesis provides insight into the relationship between ToM and behaviour in children with and without ASC, which could inform the development of well-researched and evidenced clinical interventions. It provides a valuable contribution to the relatively scarce literature on PDA traits in ASC, with important implications for the ASC assessment process. In addition, it highlights the need for individualised behavioural management strategies to help improve the quality of life for children and families.

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Part 1: Literature Review

The Relationship between Theory of Mind and
Aggression in Childhood:

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1.1 Abstract

Aims: Previous research into the relationship between theory of mind (ToM) and aggression in children does not report conclusive findings. This review aims to investigate this relationship further with a focus on potential moderating factors.

Method: A systematic literature review was conducted using PSYCinfo, MEDLINE and Pubmed. Databases were searched using terms relating to ToM and aggression and results were limited by age. This identified 148 studies, 29 of which were retrieved in full. Of these, 19 studies met the criteria for inclusion in the analysis. Correlations statistics between ToM and aggression were extracted or calculated from the reported data and a meta-analysis was conducted using a random effects model. Additional analyses were performed to assess factors influencing the relationship between ToM and aggression. Further consideration was given to the longitudinal and predictive nature of this relationship with a narrative synthesis of findings.

Results: The aggregate random effects estimate for the relationship between ToM and aggression was $r=-.10$, indicating a weak negative association between ToM and aggression. There was no significant moderating effect of age or the type of aggression measured, although the relationship was stronger for younger children. Longitudinal analyses suggest ToM may be predictive of later aggressive behaviour. However, findings were not consistent across studies and were moderated to a significant extent by other factors.

Conclusions: The review found a weak but significant relationship between better ToM and lower rates of aggression, contributing to an understanding of the link between ToM and behaviour in childhood. More research is needed to further investigate the effect of moderating variables and different types of aggression.

1.2 Introduction

1.2.1 Theory of Mind (ToM)

ToM is defined as the ability to attribute mental states to ourselves and others (Premack & Woodruff, 1978) and to explain behaviour based on the minds of others, understanding that these are different to our own (Frith & Frith, 2005). This knowledge; that emotions, beliefs and other internal experiences lead to human action, is thought to be central in developing a coherent and accurate understanding of people and their behaviour (Wellman, Cross & Watson, 2001). The development of ToM is complex and multi-faceted (Hughes & Leekam, 2004). However, it is generally accepted that this begins in early childhood, with precursors to ToM and a rudimentary understanding of other's minds thought to be present even in children as young as nine months old (Baron-Cohen, 1991; Cadinu & Kiesner, 2000).

Perhaps the most well-known test of ToM used with young children is an assessment of 'false-belief understanding'. The false belief paradigm requires a child to predict someone else's behaviour in the event of an unexpected occurrence. This draws on the understanding that another, who has been privy to different knowledge, will have different beliefs. Arguably the best known example of this is the 'Sally-Anne' task (Baron-Cohen, Leslie & Frith, 1985) whereby a character, Sally, puts an object in a box and leaves the room. Unbeknownst to her, another character, Anne, moves the object somewhere else. The child is then asked to explain where Sally will look for her object on her return, drawing on their grasp of the knowledge that Sally's beliefs and actions will be based on her own understanding, which is different from their own. Typically, success on the false belief test, and therefore evidence of basic ToM skills, is thought to begin to emerge around the age of three and is usually observable by the age of five (Perner, 1991).

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Impairments in ToM have been associated with neurodevelopmental conditions such as autism spectrum condition (ASC) and attention deficit hyperactivity disorder (ADHD) (Korkmaz, 2011). They have also been linked with diagnoses such as schizophrenia (Sprong, Schothorst, Vos, Hox & van Engeland, 2007) and major depression (Lee, Harkness, Sabbah & Jackson, 2005). However, whilst impairments in ToM are associated with neurodevelopmental conditions and mental health diagnoses, they are also evident in typically developing populations. In this group, deficits in ToM have been linked to difficulties with social behaviour (Hughes & Leekam, 2004); problems understanding the behaviour of others and the effects of one's own behaviour, and struggles with social reciprocity (Barnes-Holmes, McHugh & Barnes-Holmes, 2002; Schug, Takagishi, Benech & Okada, 2016).

These difficulties are particularly salient in childhood, where there is more likely to be greater difference in ToM capacity between individuals. However, evidence of the relationship between ToM and social behaviour is not conclusive and there is disagreement in findings (Cutting & Dunn, 1999; Frith, Happe & Siddons, 1994; Repacholi & Slaughter, 2003). A recent study has conducted a comprehensive investigation of the relationship between positive social behaviours and ToM (Imuta, Henry, Slaughter, Selcuk & Ruffman, 2016). However, at the time of writing no such review has been conducted for other, more negative behaviours and their relation to ToM.

1.2.2 Aggression

Aggression can be defined as *'feelings of anger or antipathy resulting in hostile or violent behaviour; readiness to attack or confront'* (Oxford Living Dictionaries). It can be conceptualised as a social behaviour, the aim of which is to cause harm to another individual against their wishes (Baron, 1977). This definition captures both

the more violent, physical forms of aggression, and the more subtle, indirect types of aggressive behaviour.

The development of aggressive behaviour in childhood is complex, with many factors thought to contribute to its aetiology (Reebye, 2005). Much research has studied the underpinnings of aggression, with theories ranging from biological causes (Buss & Shackelford, 1997) to behavioural ideas of reinforcement and conditioning (Patterson, Chamberlain & Reid, 1982). Other suggestions have ranged from a systemic standpoint (Dodge, 1980) to a more cognitive understanding (Dodge, 1986). In typically developing children, varying levels of aggression are expected to emerge at different stages of development (Faggot & Hagan, 1991). For example, the concept of the 'terrible twos' is based on the generally accepted idea that this stage in a child's development is associated with a notable increase in aggressive behaviours. However, more problematic expressions of aggression occur when behaviour becomes particularly extreme or is displayed outside of an expected developmental timeframe (Tremblay et al., 2004).

In developing an understanding of the construct of aggression, much research has focused on defining the different forms and functions of aggressive behaviour. Forms of aggression relate to the observable characteristics of behaviour or to the means of the behaviour being carried out. The most frequently observed distinctions here are between physical or direct aggression and relational or indirect aggression (e.g. hitting, shouting, pushing vs. gossiping, spreading rumours, socially excluding others) (Lundh, Daukantaite & Wangby-Lundh, 2014). There is also a distinction between the different functions of aggression, distinguishing between aggressive behaviours which are 'cold-blooded' or pre-planned (proactive) and those which are retaliatory and more impulsive (reactive).

Although the definitions of the forms and functions of aggression differ, there is a school of thought that argues the different types of aggression are interchangeable and hang together as one over-arching construct (Carlson, Marcus-Newhall & Miller, 1989). This is supported by research that suggests different types of aggression occur consistently across genders and that any observable differences are trivial in nature (Card & Little, 2006; Card, Stucky, Sawalini & Little, 2008). However, there is a strong counter-argument that there is a lack of association between different forms and functions of aggression and as such, the concept would be better explained by distinct multi-factored models (Poulin & Boivin, 2000). This is supported by contradictory findings that different types of aggression occur to varying degrees across genders (Lansford et al., 2012). The argument for a clear distinction between the forms and functions of aggression is of particular relevance when considering the developmental trajectory of aggressive behaviour.

There is general agreement that the trajectory of aggressive behaviour remains relatively stable over time (Huesmann, Eron, Lefowitz & Walder, 1984), with higher levels of aggression in childhood (over and above what can be classed as developmentally appropriate) acting as a predictor of more serious antisocial behaviour in adolescence and adulthood. Additionally, the stability of aggression across generations is as consistent as it is within individuals (Doumas, Margolin, & John 1994; Eron & Huesmann, 1990). This inter-generational maintenance of aggressive behaviour raises concerns as it can be damaging not only at an individual level, but to society more broadly. As such, there is a large body of research centred on understanding more about the underlying causes and related socio-cognitive correlates of aggressive behaviour (Farrington, 1991; Hubbard, McAuliffe, Morrow &

Romano, 2010), essential to facilitate the development of much needed strategies to address and prevent the development of problematic aggressive behaviour in children.

1.2.3 ToM and Aggression

To better understand aggressive behaviour, it is important to consider the relation to socio-cognitive skills and abilities (Imuta et al., 2016).

Specifically, many studies have investigated ToM and aggressive behaviour, proffering evidence to suggest there may be an association between the two (Crick & Dodge, 1996; Harvey, Fletcher & French, 2001). However, findings have differed between studies and the direction of this relationship is not conclusive (e.g. Capage & Watson, 2001; Walker, 2005). One explanation for this discrepancy in findings may be that the link between ToM and aggression is effected by the type of aggression being measured. For example, proactive and reactive aggression describe different behaviours which could be related to ToM in different ways. Children who display reactive aggression are more likely to be directly aggressive towards others. This has been associated with disorders such as conduct disorder, which in turn have been linked to impairments in ToM (Happe & Frith, 1996). Contrastingly, children who use more proactive forms of aggression, pre-meditated to facilitate personal again, e.g. bullies, may show better-developed ToM skills than their peers (Sutton, Smith & Swettenham, 1999) in order to successfully use more socially manipulative strategies.

There are also implications arising from the developmental nature of ToM. Generally, children have developed the basic groundings of ToM by the age of five. However, the development of more 'affective' ToM skills, understanding other's emotions and feelings, continues throughout childhood and into adolescence (Austin, Bondu & Elsner, 2017). In addition, there are age related differences in aggressive behaviour, with a general trend from direct to more indirect expressions of aggression

evident as children mature (Rivers & Smith, 1994). Given this, differences in the relationship between ToM and aggression may be expected to emerge as children age. However, conceptually there are a number of other variables likely to impact this relationship, including gender, language development and other co-occurring behaviours, which are difficult to control for.

As such, a complex and unclear picture of the relationship between ToM and aggressive behaviour emerges, influenced by a variety of factors including the type of aggression being measured and how this is conceptualised, and the roles of other variables such as age. Further knowledge of this relationship would likely contribute to a better understanding of factors underpinning aggressive behaviour.

1.2.4 Research Aims

Previous research has focused on understanding more about the role of ToM in behaviour, with a number of studies investigating the relationship between ToM and aggression. However, the existing literature reports contradictory findings and does not provide conclusive evidence on the direction or nature of this relationship. Further investigation of this would contribute to an increased understanding of the role of socio-cognition in aggressive behaviours. As such, this review aims:

1. To provide an overview of the relationship between ToM and aggression in typically developing children using meta-analytic methodology.

2. To consider the influence of potential moderating factors on this relationship.

3. To explore the longitudinal relationship between ToM and later aggression.

Thus, this review will inform an understanding of the relationship between ToM and aggression, with the potential to give rise to more informed preventative or targeted

therapeutic interventions. This will provide a valuable contribution to the wider literature exploring the role of social-cognitions in behaviour.

1.3 Method

1.3.1 Literature Search and Inclusion Criteria

Three electronic databases (PSYCinfo, MEDLINE and Pubmed) were used to conduct a systematic literature search. Search terms for ToM were combined with terms associated with aggression. Exact search terms are detailed in Figure 1.1. The results were limited by age to exclude studies with adult participants. In addition, informal searches were conducted by entering relevant terms into an internet search engine, Google Scholar.

Once duplicates were excluded, 148 studies remained. These studies were then screened via a two-stage process.

Phase 1. Firstly, the abstracts of articles were assessed according to the following inclusion criteria:

1. Participants were typically developing children aged two-twelve years old.

2. ToM was measured.

3. Aggression was measured.

4. Studies were empirical papers based on new data (as opposed to reviewing existing findings).

5. Studies were published in English language, in a peer reviewed journal.

6. Correlation analyses were reported.

Phase 2. Secondly, studies which appeared to meet these criteria based on their abstracts were retrieved in full for more detailed evaluation. The methodology and analyses of these studies were assessed in more detail to ensure appropriate correlation

analyses were reported. In addition, measures of ToM and aggression were assessed in accordance with the inclusion criteria described below. This resulted in the identification of 19 studies eligible for inclusion in this review. The search strategy and results are illustrated in Figure 1.1.

1.3.2 ToM Measures

Provided a task-based assessment was included, no measures of ToM were immediately excluded from the review. The most common method of assessing ToM tends to be to an aggregate score based on several false belief tasks and other language based assessments of ToM (Wellman et al., 2001). As children's performance on a variety of ToM tests has been shown to be highly correlated (Slaughter & Gopnik 1996; Watson, Nixon, Wilson & Capage, 1999), this method of testing is deemed to be appropriate and reliable.

In studies where there was a distinction between cognitive and affective ToM (n=1), the statistic for cognitive ToM was used in the meta-analysis. As the construct maps more to the types of ToM assessed by false belief tasks it was deemed to be more in keeping with overall measures of ToM reported.

1.3.3 Aggression Measures

Measures of aggression were deemed eligible for inclusion in the review provided they were described as measuring 'aggression' and were based on informant-report or observation-based measurement tools. Although there is significant overlap between the two constructs, measures of externalising behaviour alone were not included in the review as they were not felt to be fully representative of the construct of aggression (Liu, 2004).

Measures of all forms and functions of aggression were eligible for inclusion in the review. If data was reported for one form or function of aggression only, this

was used in the meta-analysis. Where more than one type of aggression was reported (n=5), only one statistic was used as multiple effects may be dependent on one another in the data. Decisions about which figure to include in the overall meta-analysis were based on information reported in the existing literature.

Where data were reported for both reactive and proactive aggression, the data for reactive aggression were used in the meta-analysis. Reactive and proactive aggression have been shown to be highly correlated, up to $\varphi = .85$, $p<.001$ (Kempes, Matthys, Maassen, van Goozen & van Engeland, 2006; Murray-Close, Ostrov, Nelson, Crick & Coccoro, 2010; Poulin & Boivin, 2000). However, reactive aggression is thought to be more stable between genders than proactive aggression (Euler, Steinlin & Stadler, 2017). Given the high correlations reported between reactive and proactive measures of aggression, this should make only minimal difference to the results.

Where indirect/relational and physical aggression were measured separately, the statistic for indirect aggression was used in the meta-analysis. Indirect aggression has been shown to significantly overlap with other forms of aggression, perhaps as there are several ways in which someone can be indirectly aggressive (Buss & Perry, 1992; O'Toole, Monks & Tsermentseli, 2017). Whilst the relationship between indirect and physical aggression is highly correlated in both boys and girls, research has also shown that significant gender differences occur in rates of physical aggression (Crick, Casas & Mosher, 1997; Vaheedi & Nazari, 2010). As indirect aggression occurs more consistently across genders (Lansford et al., 2012), these statistics were deemed to be more appropriate for use in the overall meta-analysis.

1.3.4 Longitudinal Analyses

To investigate the third aim, further review of the longitudinal and predictive relationship between ToM and later aggression was undertaken. Studies were included

in this analysis provided they reported a β or *r* statistic for the relationship between ToM at Time 1 and Aggression at Time 2. Reports of all forms and functions aggression were included. The role of the different types of aggression was considered in this relationship.

1.3.5 Data Inclusion and Calculations

1.3.5.1 Correlation coefficient calculations. Studies were only included in the meta-analysis if they reported a correlation coefficient (*r*) between ToM and aggression measured at the same time point, or contained sufficient data to allow the calculation of a correlation coefficient.

For included studies which did not report a correlation coefficient (n=2), this was calculated using the method proffered by Peterson & Brown (2005). They dictate that it is possible to compute *r* from β within a range of values (-0.5 to 0.5). They argue that this can be done using the following *'ceteris paribus'* formula (which is appropriate even when the assumption that missing *r* values would not relate to the magnitudes or directions of available *r* values cannot be made):

$$r = \beta + 0.05\lambda$$

(Where λ=1 when β is non-negative and λ=-1 when β is negative).

1.3.5.2 Statistics for longitudinal analyses. In order to examine the longitudinal relationship between ToM and later aggression, any reported *r* or β statistic comparing ToM at Time 1 with aggression at Time 2 will be used. There will be a primary focus on the direction and significance of any predictive relationships.

1.3.6 Statistical Procedures

A random effects model was used to conduct an overall meta-analysis of the data, using the 'metafor' package in R (Viechtbauer, 2010). To do this, correlation coefficients were transformed from *r* to z using Fisher's z transformation:

$$z' = .5[\ln(1+r) - \ln(1-r)]$$

The chi-squared value of the heterogeneity of effects was calculate using a Restricted Maximum Likelihood Estimator with the Q statistic (Hedges & Olkin, 1985; Viechtbauer, 2002).

Additional analyses were carried out using a mixed effects model to explore the effects of age on the data. The significance of effect was calculated using a Wald-type statistic with a chi-squared distribution in R, QM.

1.4 Results

1.4.1 Search Results

The results of the literature search and screening process are illustrated by Figure 1.1.

1.4.2 Studies Included in Review

Details of the 19 studies which met criteria for inclusion in this review are outlined in Table 1.1. In studies which reported results for more than one type of aggression, only one r statistic was used in the meta-analysis (n=3; as described above). For studies which reported results by gender (n=2), both r statistics were included in the analysis as participants represented different groups.

Studies shaded in grey (n=2) did not meet criteria for inclusion in the overall meta-analysis as they calculated correlations using only a longitudinal design (e.g. the relationship between ToM at Time 1 and aggression at Time 2). However, they, alongside other studies reporting longitudinal results (highlighted in bold), provide important contributions to understand the relationship between ToM and aggression. As such, they have been included as they help address the third aim of this review and inform a narrative synthesis of results describing the longitudinal relationship.

Search terms	Databases	Number of articles
Theory of mind: theory of mind *or* TOM	PSYCinfo	n = 78
AND	MEDLINE	n = 80
Aggression: aggress* or aggressive behaviour or aggression or violen*	Pubmed	n = 31
	Google Scholar	n = 3
Limits: Human, Children (aged 0-18), English language		

Total number of articles (duplicates removed)	**n = 148**

119 articles excluded after reading titles and abstracts because: participants were older than 12 (n=36); aggression was not measured (n=14); ToM was not measured (n=10); not investigating relevant subject matter (n=16); not an empirical study (n=33); not published in a peer reviewed journal (n=9); full text unavailable online (n=1)

29 articles retrieved in full to assess whether they met inclusion criteria

10 articles excluded for the following reasons: not TD participants (n=1); unsuitable measure of aggression (n=5)*; no correlation data for ToM and aggression (n=4)

19 articles included in final meta-analysis

* Only reported 'externalising behaviours' in place of measure of aggression (n=3); measure of aggression not based on observation or informant report (n=2).

Figure 1.1 Literature search strategy and results

1.4.3 ToM and Aggression Measures

Information regarding the measures of ToM and aggression reported by the studies included in the review can be found in Table 1.2 and Table 1.3. As is detailed

in the tables, measures of ToM and aggression differed between studies, with no two studies employing the exact same measurement tools. Measures were assessed for quality based on reported reliability and validity statistics.

Table 1.1 Corpus of studies

Study	N (M,F)	Age Range (M - months)	Sample	Aggression Type *
Austin et al. (2017)	**232 (139, 93)**	**7 (90)** **8 (99)**	**TD**	*Proactive Aggression* **Reactive Aggression**
Capage & Watson (2001)	51 (20, 31)	3 – 6 (62)	TD	Aggressive Behaviour
Choe, Lane, Grabell & Olson (2013)	**239 (118, 121)**	**3 (41)** **6 (77)**	**TD****	**Peer Aggression**
Gomez & Talwar (2015)	426 (216, 210)	6 – 9 (97)	TD	Relational Aggression *Physical Aggression*
Kokkinos, Voulgaridou, Mandrali & Parousidou (2016)	120 (51, 69)	10 – 12 (132)	TD	Relational Aggression
Korucu, Selcuk & Harma (2017)	212 (106, 106)	3 – 6 (53)	TD	Aggressive Behaviour
Lane et al. (2013)	102 (51, 51)	3 – 5 (53)	TD	Aggressive Behaviour
Longobardi, Spatari & Rossi-Arnoud (2016)	150 (77, 73)	7 – 12 (101)	TD	Aggressive Behaviour
O'Toole et al. (2017)	106 (51, 55)	3 – 6 (61)	TD	Relational Aggression *Physical Aggression*
Olson, Lopez-Duran, Lunkenheimer, Chang & Sameroff (2011)	**199 (81, 118)**	**3 (41)** **6 (77)**	**TD****	**Peer Aggression**
Pellegrini et al. (2011)	88 (45, 43)	2 – 4 (44)	TD	Aggressive Behaviour
Renouf et al. (2009)	**399 (186, 213)**	**5 (63)** **6 (72)**	**TD**	**Indirect Aggression** **Physical Aggression**
Renouf et al. (2010)	**574 (268, 306)**	**5 (64)** **6 (72)**	**TD**	**Proactive Aggression** **Reactive Aggression**
Slaughter, Dennis & Pritchard (2002)	70 (41, 39)	4 – 6 (64)	TD	Aggressive Behaviour
Song, Volling, Lane & Wellman (2016)	208 (91, 117)	2 – 4 (43)	TD	Aggressive Behaviour
Walker (2005)	111 (48, 63)	3 – 5 (50)	TD	Aggressive Behaviour
Wellman, Lane, LaBounty & Olson (2011)	146 (86, 60)	3 (42) 5 (66)	TD***	Aggressive Behaviour
Werner, Cassidy & Juliano (2006)	67 (32, 35)	3 – 5 (52)	TD	Overt Aggression
Wright & Mahfoud (2014)	82 (48, 34)	3 – 6 (62)	TD	Aggressive Behaviour

* Measures of aggression in italics NOT included in the overall meta-analysis

** TD but slight over representation of high-externalising behaviours

*** TD but deemed to be at higher risk for developing conduct disorder

Table 1.2 Measures of ToM

Study	Reported Measures of ToM
Austin et al. (2017)	Aggregate score of 0-6 for cognitive and affective ToM on a cartoon task (Sebastian et al., 2012; Völlm et al., 2006) where the correct ending to a scenario was identified.
Capage & Watson (2001)	Aggregate score of 0-4 on two false belief task adapted from previous research based on unexpected contents and switched locations.
Choe et al. (2013)	Aggregate score of 0-8 across two tasks based on standard measures of false belief prediction and explanation (Bartsch & Wellman, 1989).
Gomez & Talwar (2015)	Aggregate score of 0-6 on two false belief vignettes based on unexpected location and hidden knowledge.
Kokkinos et al. (2016)	Total scale score of 0-3 based on battery of false belief tasks including unexpected contents and hidden emotions.
Korucu et al. (2017)	Aggregate score of 0-6 using Wellman and Lui's (2004) battery of ToM tasks including false belief, unexpected contents and hidden emotion.
Lane et al. (2013)	Score of 0-2 on two false belief tasks of unexpected contents (Perner, Leekam & Wimmer, 1987) and switched location (Wimmer & Perner, 1983).
Longobardi et al. (2016)	Aggregate score of 0-5 on two second order false belief tasks comprising of a look-prediction and say-prediction (Grazzini & Ornaghi, 2012).
O'Toole et al. (2017)	Aggregate score of 0-3 on two false belief tasks based on unexpected contents and change of location.
Olson et al. (2011)	Aggregate score of 0-1 on the False Belief Prediction and Explanation Tasks (Bartsch & Wellman, 1989).
Pellegrini et al. (2011)	Aggregate score of 0-5 on two false belief tasks using unexpected contents and change in location adapted from Wimmer & Perner (1983).
Renouf et al. (2009)	Aggregate score of 0-6 on standard tests of unexpected contents (Perner, Leekam & Wimmer, 1987) and unexpected identity.
Renouf et al. (2010)	Aggregate score of 0-6 on an unexpected identity and false belief task adapted from Flavell et al. (1983).
Slaughter et al. (2002)	Aggregate score of 0-5 on five verbal measures. Two unexpected contents false belief tasks (Gopnik & Astington 1988), a conflicting emotion task, a conflicting desire task and a version of the 'four sweets' task (Baron-Cohen, 1994).
Song et al. (2016)	Aggregate score of 0-6 using Wellman & Lui's (2004) battery of ToM tasks including false belief, unexpected contents and hidden emotion.

Study	Reported Measures of ToM
Walker (2005)	Aggregate score of 0-6 on two false belief measures. One change of location task (Baron-Cohen et al., 1985) and one unexpected contents task (Perner, Frith, Leslie & Leekam, 1989).
Wellman et al. (2011)	Aggregate score of 0-6 on two switched location false belief tasks assessing predictions and explanations.
Werner et al. (2006)	Score of 0-4 based on false belief (Perner, Leekham & Wimmer, 1987) and deception (Sodian, Taylor, Harris & Perner, 1991) tasks.
Wright & Mahfoud (2014)	Aggregate score of 0-4 on four false belief tasks based on switched location and unexpected contents designed to assess 'own-other' and 'own-prior' beliefs.

Table 1.3 Measures of Aggression

Study	Reported Measures of Aggression
Austin et al. (2017)	Teacher report on the Instrument for Proactive and Reactive Aggression (IPRA; Polman et al., 2009) on a five point Likert scale.
Capage & Watson (2001)	Teacher rating on the Preschool Aggression Scale (Cohen et al., 1983) with a possible score of 0-20 and a total score above 10 indicating clinically significant aggressive behaviour (Minde, 1992).
Choe et al. (2013)	Aggregate score peer aggression based on observations and teacher report using the aggression subscale of the Caregiver/Teacher Report Form (CTRF/2-5; Achenbach, 1997).
Gomez & Talwar (2015)	Teacher report on the PSBS to rate children's relational aggression using a 5 point Likert scale.
Kokkinos et al. (2016)	Self-report on the five-item Relational aggression subscale from the Children's Social Behavior Scale-Self Report (CSBS; Crick & Grotpeter, 1995).
Korucu et al. (2017)	Aggregate score on 'play disruption' subscale of the Penn Interactive Peer Play Scale (Fantuzzo et al., 1995) and the 'anger-aggression' subscale of the Social Competence & Behaviour Evaluation (SCBE; LaFreniere & Dumas, 1996).
Lane et al. (2013)	Parent report on the Child Behaviour Checklist (CBCL/2-3; Achenbach, 1992) with a 15 item aggression subscale rated using a 3 point scale.
Longobardi et al. (2016)	Teacher report using the Indices of Social Adjustment Ability in School-Age Children (Carlo et al., 2012; Caprara, Pastorelli, Barbaranelli, & Vallone, 1992) with aggression subscale using 3 point Likert scale for each question.
O'Toole et al. (2017)	Teacher report on the 12 item Preschool Proactive and Reactive Aggression Scale (PPRA; Ostrov & Crick 2007) reporting physical and relational subscales.
Olson et al. (2011)	Composite ratings of peer aggression based on researcher observations and teacher reports (CTRF/2-5; Achenbach, 1997) at T1 and the Inventory of Peer Relations (IPR; Dodge & Coie, 1987) at T2).
Pellegrini et al. (2011)	Observation ratings using event sampling with continuous recording rules. Tallied score of frequency of aggressive events.
Renouf et al. (2009)	Teacher ratings of indirect and physical aggression using six items from the Preschool Social Behaviour Scale (PSBS; Crick et al 1997), the direct and indirect aggression scales (Bjorkqvist, Lagespetz et al., 1992) and the Preschool Behaviour Questionnaire (Tremblay et al., 1987). Scores ranged from 0-6.
Renouf et al. (2010)	Teacher ratings of proactive and reactive aggression on seven items inspired by the IPR with scores ranging between 0-6 and 0-8 respectively.

Study	Measures of Aggression
Slaughter et al. (2002)	Composite score of 0-50 based on teacher report on the aggression subscale of the CBCL (Achenbach & Edelbrock, 1983) and a behavioural questionnaire.
Song et al. (2016)	Parent report on the CBCL/1.5-5 (Achenbach & Rescorla, 2000) with a 19 item aggression subscale rated using a 3 point scale, rating an overall disposition to engage in aggressive behaviours.
Walker (2005)	Teacher report on the Profile of Peer Relations (PPR; Walker, Berthelsen & Irving, 2000 cited in Walker, 2005) with an aggressive behaviour factor score.
Wellman et al. (2011)	Parent report on the CBCL/2-3.
Werner et al. (2006)	Researcher observations of free play, coded on a system based on Ladd, Price & Hart (1988) and Pettit, Bakshi, Dodge & Coie (1990).
Wright & Mahfoud (2014)	Teacher report on one aggression item, scored on a 5 point Likert scale.

1.4.4 Study Quality

An assessment tool was devised to assess study quality and suitability as no existing tools were deemed to fully represent the assessment criteria needed for this review. This is detailed in Figure 1.2. The tool was based on the Critical Appraisal Skills Programme appraisal of cohort studies (CASP, 2014) and the National Institute of Clinical Excellence quality appraisal checklist for quantitative studies reporting correlations (NICE, 2012). The tool was a ten-item measure that assessed the quality of (A) the study population and sample, (B) the methodology, (C) the results and (D) the generalisability of findings. It employed a scoring system based on a 'traffic light' scale of red, amber and green, where red represents a lack of information or paucity of quality, amber an adequate level of quality and green a good level of quality. Study quality ratings are shown in Table 1.4. In cases where evidence could not be obtained to make an accurate rating, the assumption was made that the criteria in question had not been met, i.e. it was rated as red.

A. Population
    1. Is there enough information about the sample? Is it well described? Was it recruited in an appropriate way? Is there an appropriate age range and equal distribution of gender?
    2. Is the sample representative of the population being studied? Has consideration been paid to how sample is defined as typically developing?

B. Method
    3. Is there a clear focus? Is the research question clear? Is the need for the study clearly defined?
    4. Were the measures reliable? Is there an alpha? Have the measures been validated for use in the population? Does the measure of aggression measure what it intends to? E.g. is broad measure of aggression representative of entire range of aggressive behaviours? Are measures of ToM multi-method or multi-informant? Is there adequate description of measures in order to assess quality?
    5. Was bias considered in the results? Where measurements relied on parent or teacher-report was this discussed? Where ToM tests conducted appropriately to reduce potential bias?

C. Results
    6. Were analyses appropriate? Were correlation methods used and reported? If not, could an r statistic be derived from the data?
    7. Were results precise enough (e.g. did they report 95% confidence intervals)?
    8. Were moderating variables controlled for (age and gender specifically)? Was this adequately explained in the results section?

D. Generalisability
    9. Do you believe the results? Do the findings fit with other results in the literature? Are unexpected findings explained with reference to wider literature and methodology?
    10. Are the results relevant to this analysis specifically? Is there a correlation statistic reported for measures of ToM and aggression – are types of aggression considered?

Figure 1.2 Questions for quality appraisal tool

Table 1.4 Quality rating of studies

Study	A1	A2	B3	B4	B5	C6	C7	C8	D9	D10
Austin et al. (2017)	🟢	🟢	🟢	🟡	🟡	🟢	🟢	🟢	🟢	🟢
Capage & Watson (2001)	🟢	🟡	🟢	🟢	🟡	🟢	🟡	🟢	🟢	🟢
Choe et al. (2013)	🟢	🟡	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢
Gomez & Talwar (2015)	🟢	🟢	🟡	🟡	🟡	🟢	🟢	🟢	🟡	🟢
Kokkinos et al. (2016)	🟡	🟢	🟢	🟡	🟡	🟢	🟢	🟢	🟢	🟢
Korucu et al. (2017)	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢
Lane et al. (2013)	🟢	🟡	🟢	🟡	🟢	🟢	🟢	🟢	🟢	🟡
Longobardi et al. (2016)	🟢	🟢	🟢	🟡	🟡	🟢	🟢	🟢	🟢	🟢
O'Toole et al. (2017)	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢
Olsen et al. (2011)	🟢	🟡	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢
Pellegrini et al (2011)	🟢	🟢	🟢	🟡	🟡	🟢	🟢	🟢	🟢	🟢
Renouf et al. (2009)	🟢	🟢	🟢	🟡	🟢	🟢	🟢	🟢	🟢	🟡
Renouf et al. (2010)	🟢	🟢	🟢	🟡	🟢	🟢	🟢	🟢	🟢	🟡
Slaughter et al. (2002)	🟡	🟡	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟢
Song et al. (2006)	🟢	🟡	🟢	🟢	🟢	🟢	🟢	🟢	🟢	🟡
Walker (2005)	🟡	🟢	🟢	🟡	🟡	🟢	🟢	🟢	🔴	🟢
Wellman et al. (2011)	🟢	🟡	🟡	🟢	🟢	🟢	🟢	🟡	🟡	🟡
Werner et al. (2006)	🟢	🟢	🟢	🟢	🟢	🟡	🟡	🟢	🟡	🟢
Wright & Mahfoud (2014)	🟢	🟡	🟡	🔴	🟡	🟢	🟢	🟢	🟡	🟡

1.4.5 Meta-Analysis Results

To test the first aim, a meta-analysis was conducted to investigate the relationship between ToM and aggression. A forest plot of the analysis is shown in Figure 1.3. The results show that the aggregate random effects model estimate for the relationship between ToM and aggression is r=-.10. This is a weak but significant negative relationship (z=-2.61, p=<.01 [95% CI = -.18 to -.03]) (Cohen, 1977), signifying children with lower ToM are rated as demonstrating more aggressive

behaviours. Further analysis revealed 71.38% of the variation reflected actual differences in the population mean.

Although only a small effect, just five studies reported positive correlations contradicting the overall negative relationship. Of these five, it is worth considering the quality appraisal ratings. One figure represents a male only statistic (Walker, 2005 A) and one a female only statistic (Werner et al., 2006 B). Both of these studies reported small sample sizes when separated by gender, which raises the issue of how reliably the findings can be generalised across a wider population. Another used a single item un-validated measure of aggression (Wright & Mahfoud, 2014), bringing the validity of this result into question. In addition, the remaining two studies reported non-significant correlations in their own analyses with wide-reaching confidence intervals spanning both negative and positive correlations (Gomez & Talwar, 2015; Song et al., 2016). These findings should therefore be interpreted with caution.

In addition, there was significant heterogeneity between studies (Q(18)=61.21, $p<.01$). This indicates that the variability between reported z correlations is larger than would be expected from sampling error only (Lipsey & Wilson, 2001). A Baujat plot (Baujat, Mahe, Pignon & Hill, 2002) identified only one study contributing to the overall heterogeneity (Walker, 2005 A). This is of note as it was one of only two studies to report analyses for a male-only sample. However, further analysis in R (Viechtbauer & Cheung, 2010) did not reveal this to be a significant influencer on the overall analysis (due to the small sample size of the group) and as such, this data point was not removed.

1.4.6 Publication Bias

A funnel plot shown in Figure 1.4 was used to illustrate publication bias. Although this did not depict a conclusive degree of symmetry, neither Egger's

regression test (Egger, Davey-Smith, Schneider & Minder, 1997) nor the Rank

correlation test were statistically significant (*z*=.02, *p*=.98; *τ*=.02, *p*=.95 respectively).

This suggests there was no evidence of publication bias in the data.

Author(s), Year		Correlations [95% CI]	
Austin et al., 2017		-.02	[-.15 to .11]
Capage & Watson, 2001		-.28	[-.52 to -.00]
Choe et al., 2013		-.20	[-.32 to -.08]
Gomez & Talwar, 2015		.07	[-.03 to .17]
Kokkinos et al., 2016		-.18	[-.34 to -.01]
Korucu et al., 2017		-.27	[-.39 to -.14]
Lane et al., 2013		-.29	[-.53 to -.00]
Longobardi et al., 2016		-.09	[-.25 to .07]
Olson et al., 2011		-.20	[-.33 to -.06]
O'Toole et al., 2017		-.06	[-.25 to .13]
Pellegrini et al., 2011		-.19	[-.38 to .02]
Slaughter et al., 2002		-.11	[-.31 to .10]
Song et al., 2016		.04	[-.10 to .18]
Walker, 2005 A		.46	[.20 to .66]
Walker, 2005 B		-.16	[-.39 to .09]
Wellman et al., 2011		-.26	[-.41 to -.10]
Werner et al., 2006 A		-.37	[-.63 to -.04]
Werner et al., 2006 B		.05	[-.30 to .39]
Wright & Mahfoud, 2014		.13	[-.09 to .34]
RE Model		-.10	[-.18 to -.03]

-0.6   -0.4   -0.2   0.0   0.2   0.4   0.6

Correlation Coefficient

Figure 1.3 Forest plot of the random effects model meta-analysis between ToM and

aggression.
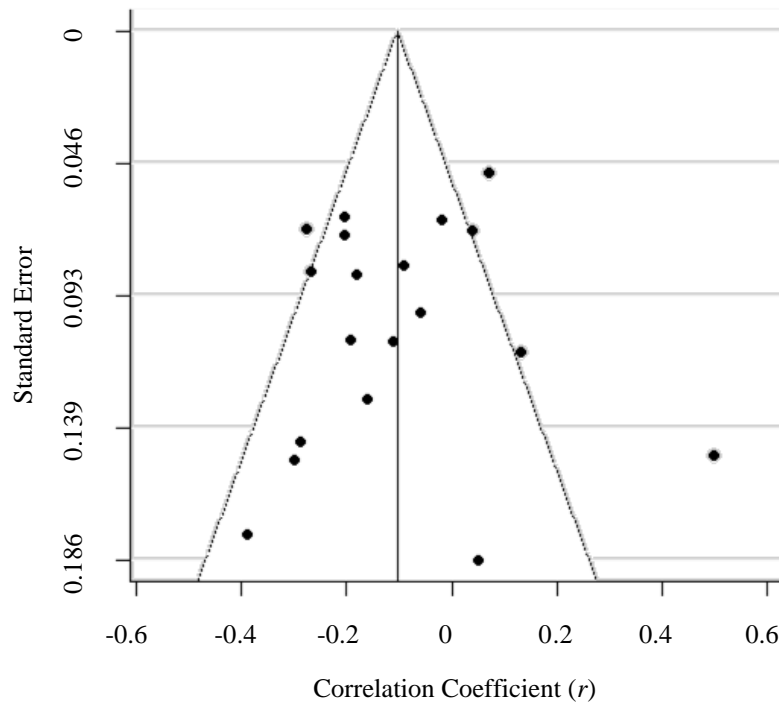
Figure 1.4 Funnel plot depicting publication bias in the studies.

1.4.7 Moderating Factors

To explore the second aim, moderating influences on the relationship between ToM and aggression were investigated using both categorical and continuous analyses. Categorical approaches are justified for use in analyses of fewer than 20 test statistics (e.g. Wykes et al., 2011). This is because there is often insufficient power to infer valid conclusions from a meta-regression.

1.4.7.1 Participant age. The effect of participant age on the relationship between ToM and aggression was tested as a continuous moderator. ToM capacity changes dramatically during childhood (Wellman, Cross & Watson, 2001), as do typical expressions of aggression (Hay, Payne & Chadwick, 2004). As the ages of participants represented the entire spectrum of childhood (between two-twelve years old), there was expected to be some moderating effect of age. The mean participant

age in months was extracted from the studies. For studies where mean age was not reported, this was calculated using an estimate derived from the range and median. Age was not found to significantly moderate the relationship between ToM and aggression (QM(1)=.36, p=0.55 [95% CI=-.00 to .00]), suggesting age did not have a significant effect on the relationship between ToM and aggression.

In addition, the effect of age was explored using categorical analysis. This allowed for additional data from studies reporting correlations at more than one age point to be included. Studies were separated into two groups by participant age, ≤five and ≥six. Age five was chosen as the categorical separation point as it is expected most children will have developed basic false-belief ToM ability by this time (Perner, 1991). The random effects model for younger children indicated a stronger effect (r=-.12, z=-2.41, p<.05 [95% CI=-.22 to .02]) than the non-significant effect reported for older children (r=-.06, z=-1.60, p=.11 [95% CI=-.14 to .01]).

1.4.7.2 Type of aggression. To investigate potential effects of the form of aggression being measured, pooled effect sizes were calculated for the different forms of aggression which were reported by two or more studies, documented in Table 1.5.

The analysis was largely in line with findings from the overall meta-analysis, suggesting there is a weak negative correlation between ToM and aggression, regardless of the type of aggression being measured. There was some variation between the strength of the relationship for the different forms of aggression, perhaps indicative of the strongest relationship occurring between peer aggression and ToM. However, given the small number of studies analysed for each form of aggression, the analysis is unlikely to have significant power to detect effects with any certainty.

Table 1.5 Pooled effect sizes for types of aggression

Type of Aggression	N of studies	N	Pooled Effect Size (*r*)	95% CI
Overall Aggression	10	1179	-0.10	-0.23 to 0.03
Peer Aggression	2	438	-0.20	-0.30 to -0.11
Relational/Indirect Aggression	3	657	-0.04	-0.20 to 0.11
Physical Aggression	2	517	-0.11	-0.32 to 0.10

1.4.7.3 Sample characteristics. Although taken from a typically developing population, three of the studies in this analysis reported data for children who were deemed to be at slightly increased risk of conduct disorder, or who showed marginally elevated rates of externalising behaviour based on parent-report measures. A meta-analysis on these studies alone reported an aggregate random effects estimate of the relationship between ToM and aggression was *r*=-.22 (*z*=-5.22, *p*<.001 [95% CI = -.30 to -.14]). This indicates a stronger relationship than was found for studies where children were not deemed to show increased rates of these behaviours (*r*=-.08, *z*=-1.66, *p*=.09 [95% CI = -.17 to .01]).

1.4.7.4 Gender. Many of the studies included in the meta-analysis reported on the effect of gender with varying results. Two studies reported gender was a significant predictor of aggression, and that boys demonstrated significantly more aggressive behaviours than girls (Kokkinos et al., 2016; O'Toole et al., 2017). Two further studies found no significant effect of gender overall (Gomez & Talwar, 2005; Werner et al., 2006). However, they found that gender was significantly related to ToM scores in younger children, with better ToM scores reported for girls at a younger age. The significance of this effect decreased as children aged. However, one additional study

also report a non-significant effect of gender for younger children in their analyses (Pellegrini et al., 2011).

Two studies, (Walker, 2005; Werner et al., 2006) conducted separate analyses by gender. Walker (2005) found significant differences between boys and girls, suggesting girls have better ToM than boys, and that boys were more aggressive than girls. Although no direct moderating effects of gender were tested for, better ToM in boys was found to be moderately correlated with higher levels of aggression, whereas better ToM in girls was weakly correlated with lower levels of aggression. Although studying children from the same age range, Werner et al. (2006) did not replicate these findings, reporting no differences in ToM between boys and girls. In direct contrast to Walker's findings, they also found aggression in boys correlated negatively with ToM, whereas this was not the case for girls.

Another notable finding was a significant interaction effect between gender and ToM on measures of relational aggression (Kokkinos et al., 2016). ToM was reported to be a significant predictor of relational aggression for girls, with better ToM related to lower levels of aggression. However, this was not the case for boys, where ToM did not significantly predict aggression.

1.4.8 Longitudinal Analyses

As the numbers of studies relevant to the third aim were low (n=5) and reported on different forms and functions of aggression, a narrative synthesis of results was conducted to investigate the longitudinal relationship between ToM and later aggression. Two studies (Renouf et al., 2009; Renouf et al., 2010), were not included in the overall meta-analysis as they employed a solely longitudinal design measuring the predictive relationship between ToM and later aggression. Given this, it was not methodologically feasible to derive an r statistic as data from cross-sectional and

longitudinal study designs could not be combined. However, these studies were methodologically rigorous with large sample sizes. Given this, the potential contribution to this review was such that their inclusion was justified.

1.4.8.1 Indirect aggression. Renouf et al. (2009) studied the relationship between ToM at age five and indirect aggression a year later. They found ToM to be a unique and significant predictor of indirect aggression (β=.15, p<.01), with better ToM related to increased rates of indirect aggression. However, this relationship was moderated to a significant level by prosocial behaviour (β=-.11, p<.01). For children with low and average levels of prosocial behaviour, better ToM was significantly related to higher levels of indirect aggression (β=.27, p<.01; β=.15, p<.01 respectively). Contrastingly, for children with high rates of prosocial behaviour, ToM was not associated with indirect aggression.

1.4.8.2 Peer aggression. Choe et al. (2013) and Olson et al. (2011) investigated the relationship between ToM at age three and peer aggression two to three years later. Although both studies reported a very weak negative correlation between ToM and later levels of peer aggression, neither finding was significant (r=-.08, p>.05 and r=-.09, p>.05 respectively) and as such, regression analyses were not reported.

1.4.8.3 Reactive aggression. Renouf et al. (2010) studied the relationship between ToM at age five and reactive aggression one year later. They found no main effect of ToM, although a significant interaction of ToM and peer victimisation was observed (β=-.07, p<.05). In children who faced high or average levels of peer victimisation, poorer ToM was significantly related to higher rates of reactive aggression (β=-.16, p<.01 and β=-.09, p<.05 respectively). In contrast, ToM was not associated with reactive aggression in children experiencing low levels of peer victimisation.

In addition to these studies, Austin et al. (2017) explored the longitudinal relationship between cognitive and affective ToM and reactive aggression, finding that cognitive ToM at Time 1 (children aged between six - eight) significantly predicted reactive aggression at Time 2, one year later (Wald(1)=-.23, *p*<.05). They also found affective ToM at Time 1 to be a significant predictor of reactive aggression at Time 2 (Wald(1)=-.27, *p*<.01). Both findings refer to a negative relationship, with better ToM predicting lower rates of aggression later in childhood.

1.4.8.4 Proactive aggression. Renouf et al. (2010) investigated the effect of ToM at Time 1 (aged five) on rates of proactive aggression at Time 2, one year later. ToM made a unique and significant contribution (β=.07, *p*<.05), with better ToM predicting increased levels of proactive aggression. Again, this relationship was moderated to a significant extent by peer victimisation (β=.10, *p*<.01), in that ToM was only positively related to proactive aggression for children experiencing high levels of peer victimisation (β=.16, *p*<.01).

Austin et al. (2017) presented contradictory evidence, suggesting that affective ToM at Time 1 significantly predicted rates of proactive aggression at Time 2, one year later (Wald(1)=-.26, *p*<.05), with better ToM predictive of lower rates of aggression. Additionally, their analyses also found cognitive ToM not to be a significant predictor of proactive aggression, indicating further complexity in the relationship between the two variables.

1.4.8.5 Physical aggression. Renouf et al. (2009) did not find any significant predictive relationship between ToM and physical aggression.

1.4.8.6 Conclusions on narrative review. There is a general consensus that ToM is, to some level, predictive of later aggressive behaviour. However, the direction and strength of this relationship differs across studies and based on the form or function

of aggression being measured. ToM was not found to be a significant predictor of either physical or peer aggression. Better ToM was found to predict increased rates of indirect aggression one year later, but this finding was based on one study alone. There was more of a consensus that better ToM predicts lower levels of reactive aggression. However, the strength of this association differed, with some findings suggesting the significance in relationship may be dependent on the moderating effect of peer victimisation. The findings for proactive aggression were more contradictory, with studies reporting ToM to be a significant predictor in both negative and positive directions, with some influence of moderating factors.

Finally, overall predictive models, inclusive of moderating factors, appeared to account for very little of the variance observed in scores, at approximately 20%.

1.5 Discussion

Much research has explored the relationship between socio-cognitive skills and behaviour in childhood. There is general agreement that there are significant links between the two, albeit complex and multi-faceted in nature (Monks, Smith & Swettenham, 2005). Previous studies have specifically investigated the relationship between ToM and aggressive behaviour in childhood. Although most conclude there is likely to be an association between ToM and aggression, findings vary and there is a lack of consensus on the strength and direction of the relationship. This review aimed to synthesise the literature and provide evidence to inform a more comprehensive understanding of the relationship between ToM and aggression in childhood.

A meta-analysis of correlation coefficients using a random effects model revealed a weak negative relationship between ToM and aggression, such that better ToM is weakly correlated with lower levels of aggressive behaviour in typically

developing children between the ages of two and twelve. This relationship was not moderated by age, although there was increased strength in the model for younger children. The model lacked significance for children over six, perhaps in part as a result of the decreased variability in ToM in older children. In addition, the overall model was stronger for children displaying increased levels of externalising behaviour and those at elevated risk of conduct disorder, and lacked significance when these findings were removed from the analysis. The relationship between ToM and aggression varied slightly depending on the type of aggression measured. However, given the small number of studies reporting data for each type of aggression, it is unlikely there was sufficient power to detect small effects in these analyses.

Analysis of the longitudinal relationship between ToM and aggression revealed a complex picture with disparate findings. ToM did not emerge as a significant predictor of either physical or peer aggression. In keeping with the results of the meta-analysis, better ToM was predictive of lower levels of reactive aggression in later childhood. However, this relationship was subject to moderation effects, only remaining significant for children who experienced above average levels of peer victimisation. In contrast to the results of the meta-analysis, better ToM was found to predict higher levels of indirect and proactive aggression in later life, although this finding was not consistent across studies and was moderated by other social and behavioural factors. This highlights a difficulty in trying to understand the relationship between ToM and aggression in isolation, when they are both constructs which are highly related to other socio-cognitions and behaviours (Hughes & Leekam, 2004; Romano, Tremblay, Boulerice & Swisher, 2003). Further research is needed to better understand these findings.

Overall, the finding that better ToM relates to lower levels of aggression was as expected. However, there must be further consideration of factors which may moderate this relationship, and of the methodological limitations of the studies reviewed.

1.5.1 Effects of Age

Previous studies have hypothesised that, given the developmental nature of ToM and aggressive behaviour (Loeber & Hay, 1997; Wellman, 1992), age is likely to moderate the relationship between the two in some way. Whilst age did not significantly moderate the relationship between ToM and aggression in the overall meta-analysis, categorical analyses revealed the overall model was stronger in children under five years old than for older children. As evidence of basic ToM abilities tends to emerge around the age of five (Perner, 1991), one could hypothesise that this allows children to better understand the effects of their actions on others and so become less likely to engage in aggressive behaviours. This may explain why the relationship between ToM and aggression seems to become less relevant in children over five, as the development of basic ToM capacity may serve to ameliorate more aggressive behaviour.

In addition, synthesis of the longitudinal studies found ToM was positively related to proactive aggression in younger children, and negatively for older children. In older children only affective ToM, involving more complex understandings of others' feelings and emotions, was found to significantly predict proactive aggression. Many argue that whilst basic ToM is intact relatively early in life, children continue to develop skills in perspective taking and understanding throughout their development and into early adulthood (Dumontheil, Apperly & Blakemore, 2010). It may be that in younger children, proactive aggression is reliant on the more basic, cognitive ToM

abilities which allow for manipulative and pre-planned aggressive behaviours. However, when older children develop more affective ToM, a better understanding of consequence and emotions emerges. This may result in lower levels of aggressive behaviour, as the negative impact of behaviour on others is fully understood, thus providing a potential explanation for the changing nature of the relationship.

1.5.2 Type of Aggression and ToM

There is a lack of consistency in the relationship between ToM and aggression, with previous findings seeming to differ depending on the type of aggression being measured (e.g. O'Toole et al., 2017; Renouf et al., 2009). Due to power constraints, this review did not have the scope to include the type of aggression measured as a moderator in the meta-analysis. However, interpretation of pooled effect sizes did not provide sufficient evidence to suggest significant differences in the relationships between ToM and different forms of aggression.

One explanation for this finding is that most studies included in the analysis measured one, overall form of aggression, with only a smaller number measuring separate forms. This may have resulted in insufficient power to detect effects. In addition, given that different forms of aggression have been demonstrated to be highly correlated and there is an argument for them to be incorporated under the same conceptual framework (Archer & Coyne, 2005), it may be that the different forms of aggression measured by the studies in this analysis overlap significantly in mapping to the same construct. With this in mind, the findings add support to arguments for conceptualising aggression under one overarching framework. Further research centring on the profiles of different types of aggression is needed to provide weight for this argument.

When considering the effect of the forms and functions of aggression on the longitudinal relationship between ToM and aggression, there were more notable differences between findings. Some studies found better ToM was predictive of lower levels of both reactive and proactive aggression in later childhood (Austin et al., 2017), whilst others reported better ToM was related to higher levels of proactive aggression (Renouf et al., 2010). This discrepancy in findings may, in part, relate to the moderating effects of social or individual trait factors. For example, one study found peer victimisation significantly moderated the relationship between ToM and proactive and reactive aggression (Renouf et al., 2010). A potential explanation for this may be that aggressive children are perhaps more likely to experience social exclusion and victimisation, giving them less opportunity to practice their ToM skills. This means they may engage in more proactively aggressive behaviour, e.g. manipulation, in response to their experiences of victimisation. Conversely, children with better ToM may be less likely to be excluded socially and therefore have more opportunities to develop skills and cognitions which make future reactive aggressive behaviour even less likely (e.g. emotion regulation). This demonstrates how it is difficult to fully understand the relationship between ToM and different types of aggression without some consideration of moderating factors.

1.5.3 Effects of Sample Characteristics

In addition, the behavioural characteristics of the sample had a significant impact on the relationship between ToM and aggression. All studies reported data for children who were deemed as typically developing. However, three studies reported their samples to be at slightly increased risk of conduct disorder, or display marginally higher rates of externalising behaviours. The relationship between ToM and aggression was stronger for children showing these behavioural characteristics than

for those who did not, although due to the small number of studies there was limited power to detect effects. In addition, when these studies were removed from the overall analysis, the relationship between ToM and aggression no longer reached significance. This difference in association may potentially be explained with reference to the finding that children with conduct disorder typically show widespread ToM impairments (Happe & Frith, 1996). It therefore follows that we would expect children at higher risk of conduct disorder to show impaired ToM and higher rates of aggressive behaviour compared to controls, thus resulting in increased strength in the relationship. In addition, the finding that the relationship between ToM and aggression does not reach significance when these children are removed from the model raises important questions about whether there is any association between ToM and aggression in children who can truly be classified as typically developing.

However, this finding should be interpreted with caution. Although parents of the children in these samples reported an over-representation of these traits, the sample still came from a typically developing population. With this in mind, perhaps the most appropriate conclusion to be drawn from the findings is that the relationship between ToM and aggression becomes more salient as aggressive behaviour increases. It is also important to note that just because similar behavioural characteristics were not reported for many of the other samples, this does not mean they were fully representative of a typically developing population. This highlights wider issues with how samples are described and how possible it is to study a group of children whose behaviour is completely in line with reported norms.

1.5.4 Effects of Gender

Another factor implicated in the relationship between ToM and aggression is that of gender. There is a large body of existing research exploring gender differences

in both aggression and ToM, with most authors agreeing there are differences between boys and girls. Generally, boys are seen as more aggressive (Endendijik et al., 2017), while girls are though to develop ToM skills at a younger age (Walker, 2005), implying there is likely to be a role of gender in the relationship between ToM and aggression.

Overall, the results of the systematic analyses support previous findings; that girls develop ToM skills at a younger age and that boys display more aggressive behaviours. However, when interpreting this finding it is important to note that some measures of aggression are thought to contain an over-representation of items tapping one form of aggression (generally physical aggression), potentially reducing sensitivity in analyses (Card & Little, 2006; Little, Heinrich, Jones & Hawley, 2003; Renouf et al., 2010). It then follows that measures may be more sensitive to the 'male' characteristics of aggression, exposing the results to bias and over-inflating the impact of gender (Bowie, 2007). Although for the most part, studies included in this review took measures to ensure this was not the case, it is still possible that a lack of sensitivity in measurement led to an under-report of aggression in girls, biasing the findings.

In addition, some studies in this review reported findings by gender. There was some discrepancy in findings between studies. Walker (2005) found that whilst ToM is negatively related to aggression in girls, it is predictive of higher levels of aggression in boys. Conversely, Werner et al. (2006) report that aggression was associated with lower ToM in boys but not girls. In addition, Kokkinos et al. (2016) found there was a significant negative relationship between ToM and relational aggression in girls, which was not replicated in boys. This suggests there is likely to be an effect of gender on the relationship between ToM and aggression. Whilst limited findings in the review were reported by gender, it may be that more conclusive findings on the effects of gender would have emerged from the analysis of more data. Further research is needed

to fully understand the nature of these differences and to definitively conclude whether there is any effect of gender.

1.5.5 Additional Factors

Whilst possible factors influencing the relationship between aggression and ToM in this analysis have been discussed, there was a high proportion of the variance in the overall model that these factors could not account for. It is therefore difficult to see how the relationship between ToM and aggression can be understood in isolation from other known correlates of the two constructs. For example, developmental factors such as language ability have been shown to be significantly related to ToM (e.g. Longobardi et al., 2016; Slaughter et al., 2002), environmental factors such as parenting style and corporal punishment have been shown to be related to aggression (e.g. Olson et al., 2011), and behavioural or personality characteristics have been shown to relate to both ToM and aggression (e.g. Lane et al., 2013; Renouf et al., 2009; Wellman et al., 2011). Without exploring each of these correlates in further detail, alongside others not mentioned, it is unlikely that the complexity of the relationship between ToM and aggression can be understood fully.

Whilst it was not within the scope of this review to examine the effects of all other variables, further research should consider alternative explanations for this variance. However, even without a deeper knowledge of potential influencing factors, the results from this review are still helpful in understanding the nature of the relationship between ToM and aggression. This is key to informing necessary behavioural management strategies and preventative approaches to manage damaging levels of aggressive behaviour at an individual and societal level.

1.5.6 Role of Methodology

In drawing conclusions on the relationship between ToM and aggression, it is key to consider the impact of methodology, not only of this review but of the studies included.

Firstly, there is an important question regarding the validity of using meta-analytic analyses with correlational data as reported in the studies. The correlation coefficient represents the strength of association between two inherently continuous variables. Whilst the studies in this review used continuous methods to assess both ToM and aggressive behaviour, some measurements of ToM were taken on very small scales, e.g. 0-2. Although ToM is not binary and can be measured as a continuous variable (Lerner, Hutchins & Prelock, 2011), there is very little room for any difference between scores on such small scales. This may have resulted in an increased risk of type II errors in the data, implying that the method of measurement could have resulted in a weaker overall model than would perhaps have been observed otherwise.

Secondly, it must also be considered that ToM and aggression were mostly assessed using different measurement tools. Whilst there is generally a high level of reliability between measures of ToM, the variability between assessment tools is still problematic. This was especially true for measures of aggression, where there was vast variability in the forms and functions measured, and the measurement tools used to do so. When considered in line with evidence that all aggressive behaviour maps to the same over-arching construct (Archer & Coyne, 2005; Lansford et al., 2012), it is arguably justifiable to compare different forms and functions of aggression with one another. However, there is another school of thought which suggests that multiple factor models better explain aggression and that it is too simplistic not to distinguish between them (Dodge & Coie, 1987). This finding suggests comparison across different types of aggression and measurement tools may be problematic. If analyses

are not comparing like with like, it follows that there will be limited validity and reliability in the results.

In addition, there are problems with using a single informant measurement tool to assess complex behavioural constructs. The studies in this review primarily relied on the use of teacher-report tools to measure aggression. Whilst teachers are seen as valid and reliable informants for measuring observable behaviours evident in school, such as aggression (Barker et al., 2008), they may be less aware of the more subtle, proactive types of aggression. This could have resulted in biased reports of aggression, potentially limiting the understanding of how the type of aggression measured effects the relationship between ToM and aggression. In addition, teacher-report measures may have reduced reliability as they depend on respondent's observational skills in the classroom which can vary between individuals. A multi-informant approach may have improved the validity of the results and yielded higher effect sizes.

It is also worth noting that when considering the clinical relevance of the findings, relying on correlational analyses means there is no way of establishing a cause and effect relationship between variables.

1.5.7 Defining Constructs

It is also important to consider the impact of the inclusion criteria specified for this review on the outcome. As the wider literature on socio-cognitions and behaviour has such brevity, it was difficult to decide on search criteria which captured all relevant studies, without returning an unmanageable number of results. Whilst narrowing the search criteria undoubtedly facilitated the identification of studies appropriate for meta-analysis and increased the reliability of the search strategy, understanding the full implications of the relationship between ToM and aggression may have been better facilitated by widening the search criteria to include related terms.

Specifically, by focusing solely on a well-defined construct, ToM, studies which looked at related socio-cognitive constructs were not captured. For example, there is a large body of research investigating the relationship between empathy and aggressive behaviour in childhood (e.g. Vachon, Lynam & Johnson, 2014) which could have provided a valuable contribution in understanding the broader relationship between socio-cognitions and aggression. Additionally, the search terms only captured specific measures of aggression. Many studies in the literature have measured concepts or behaviours such as externalising behaviour, bullying, or peer victimisation in childhood, which were not included in the review. This is because it was felt these concepts or behaviours were not completely representative of the construct of aggression. However, inclusion would likely have been helpful to further understand the relationship between ToM and aggressive behaviour. In particular, there is a wealth of literature which examines bullying, aggressive behaviour and empathy or affective ToM which may have facilitated a deeper understanding.

There must also be some consideration of the differences between 'cognitive' and 'affective' ToM. There is an argument based on behavioural and neuroimaging evidence that ToM should be separated into cognitive (concerned with beliefs and cognitive perspective taking abilities), and affective (concerned more with emotional understanding and empathy) components (Hein & Singer, 2008; Shamay-Tsoory & Aharon-Peretz, 2007). There is existing research which suggests that affective and cognitive ToM differ in their relation to behaviour and presentation, for example, the finding that affective ToM alone may predict aggression in children with callous-unemotional traits (Stellwagen & Kerig, 2013). With this is mind, it may be that different forms of ToM relate differently to the different types of aggression. As the

studies included in this review did not allow an exploration of this distinction, empirical research in this area would likely further inform understanding.

1.5.8 Clinical Implications

Although there are limitations to the conclusions which can be drawn from the findings of this review, there are important clinical implications.

Understanding more about the cognitive factors underlying aggressive behaviour may be helpful in supporting the development of effective strategies to ameliorate the negative effects of the behaviour. Aggression remains relatively stable throughout the lifespan (Huesmann & Eron, 1984; Piquero, Carriaga, Diamond, Kazemian & Farrington, 2012) and can have a hugely detrimental impact at an individual and wider societal level. Thus, clinical interventions focusing on reducing aggressive behaviours in childhood should be a key priority for research. As the findings from this review suggest better ToM is only weakly correlated with lower levels of aggression, there is not sufficient evidence to suggest that interventions aimed at supporting the development of ToM skills would have any impact on rates of aggressive behaviour. Further research is needed to explore the underlying mechanisms behind aggression in childhood in order to ameliorate the more damaging effects of aggressive behaviour.

In addition, given the correlational nature of this review, further research is needed to establish the existence of any cause and effect relationship.

1.5.9 Conclusions

In conclusion, this review contributes further insight into the understanding of the relationship between ToM and aggressive behaviour in typically developing children. It demonstrates there is a weak but significant relationship between better ToM and lower levels of aggression in childhood, which is stronger for younger

children and weaker for children aged six and over. It is unlikely that the different forms or functions of aggression measured had any significant effect on this relationship overall, although this may, in part, relate to the methodology of the review. In addition, the relationship was stronger for children who displayed increased rates of aggressive behaviour, although it is difficult to draw definitive conclusions from this finding due to the small number of studies analysed.

With regards to the longitudinal relationship, findings were not consistent across studies. Most agreed that, generally, better ToM earlier in childhood was predictive of less aggressive behaviour later in childhood. However, a significant proportion of the variance in aggressive behaviour could not be explained by ToM. Thus, the relationship is likely to be complex and dependent on a multitude of other factors, into which further research is needed.

In the context of the limitations considered, it is not possible to draw more definitive conclusions from the findings of this review. There is a notable discrepancy between measures and approaches which conceptualise aggression as a single-factor model, and those which suggest a multi-factor model. There are also difficulties with the feasibility of studying the relationship between ToM and aggression in isolation from the vast number of correlating variables. Moving forward, efforts should be made to further explore these factors to facilitate a broader understanding of the relationship between socio-cognitions and behaviour. This would add brevity and depth to the understanding of the relationship between ToM and aggression.

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Part 2: Empirical Paper

Pathological Demand Avoidance Traits in Autism
Spectrum Condition: Their Relationship to Parent-
Reported Theory of Mind

2.1 Abstract

Aims: Pathological demand avoidance (PDA) is a term used to describe a set of traits and behaviours observed in autism spectrum condition (ASC), about which relatively little is understood. Theory of mind (ToM) is a socio-cognitive construct thought to underpin the core symptoms of ASC. This study aimed to contribute to an increased understanding of PDA behaviours in ASC by investigating the relationship between ToM and the severity of PDA and ASC traits in children with ASC. **Method:** The parents of 61 children with ASC and 30 non-ASC controls aged 6-11 completed questionnaires measuring core ASC traits, PDA traits and behaviours, and ToM. The relationship between ToM and (1) PDA traits and (2) core ASC traits was investigated in the ASC sample using correlation analyses. Between-group analyses were also conducted for children with high and low PDA traits in ASC.

Results: There was a significant relationship between core ASC traits and ToM in children with ASC, with better ToM associated with lower levels of ASC traits. Overall, ToM accounted for between 13-20% of the variance in core ASC traits. There was no significant relationship between PDA traits and ToM in children with ASC. Between-group analyses revealed significant differences in the behavioural profile of autistic children with high and low PDA traits.

Conclusions: ToM was not found to relate to PDA traits in ASC, implying that there are at least partially distinct mechanisms underlying these behaviours compared to core ASC traits. Findings from between-group analyses support previous studies; that PDA refers to a distinct set of symptoms in ASC. Further research is needed to inform an understanding of the socio-cognitive processes underpinning PDA traits in ASC.

2.2 Introduction

2.2.1 Autism Spectrum Condition (ASC)

The diagnostic and statistical manual, fifth edition (DSM-5) states that for an individual to meet criteria for a diagnosis of ASC they must demonstrate 'persistent deficits in social communication and social interaction across multiple contexts' and 'restricted, repetitive patterns of behaviour, interests or activities (RRBIs)'. These core ASC traits must be present in the early developmental stages and cause 'clinically significant impairment in areas of current functioning' (DSM-5, American Psychiatric Association, 2013). Such polythetic diagnostic criteria capture a broad variation in symptomology, evident in the significant heterogeneity observed in presentations of ASC (Betancur, 2011; Ring, Woobdury-Smith, Watson, Wheelwright & Baron-Cohen, 2008; Volkmar & Reichow, 2013). However, it is generally agreed that all ASC presentations can be broadly characterised by some degree of social impairment (Wing, 1991).

In recent years, an increase in heterogeneity in presentations of ASC has been observed (Matson & Kozlowski, 2011). This is likely related to a broadening in the definition of what constitutes ASC (Gernsbacher, Dawson & Goldsmith, 2005), reflected by the move in DSM-5 to categorise the condition as occurring on a spectrum, and to incorporate Asperger's Syndrome and Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS) (Kim et al., 2011; McPartland, Reichow & Volkmar, 2012). Under this conceptual backdrop, it has been acknowledged that not all children presenting with impairments in social communication and RRBI's seem to 'fit' the general pattern associated with the typical ASC profile (e.g. Eisenberg & Kanner, 1956; Kanner, 1943). Whilst children with more atypical presentations may reach criteria for ASC, diagnosis can be controversial, and many argue that the face validity

in diagnostic and statistical manuals is not adequate in these cases (Green et al., 2018; O'Nions et al., 2016).

This controversy has in part, been addressed by the recent move to allow for comorbid diagnoses in ASC (Smith & Matson, 2010). It is now agreed that there are high rates of comorbidity between ASC and (1) mental health conditions and (2) other neurodevelopmental disorders (Matson & Williams, 2013), which can help understand more diverse presentations of ASC. Although there is often overlap in symptomology in these cases, the same symptoms do not necessarily occur as result of the same underlying processes. For example, deficits in attention are common in both ASC and ADHD but may be attributed to different cognitive mechanisms (Bramham et al., 2009), potentially representing a more disorder specific route to behaviours. This concept of 'equifinality'; that the same behaviour may occur as a result of different processes or mechanisms (Cicchetti & Rogosch, 1996), adds to the complexity of understanding more atypical behaviours in ASC (Lenroot & Yeung, 2013).

2.2.2 Pathological Demand Avoidance (PDA)

In recent years, there has been particular interest in understanding non-compliance in ASC (O'Nions, Happe, Evers, Boonen & Noens, 2018). Extreme non-compliance in everyday situations is seen as a problematic behaviour in ASC and is categorised as an important target for treatment (Chowdhury et al., 2010). Whilst findings report that everyday non-compliance occurs generally in ASC, non-compliance with everyday requests was also described as the defining feature of PDA (Newson, Marechal & David, 2003). PDA is a term coined originally to explain a set of challenging behaviours in ASC, over and above those encapsulated by existing diagnostic criteria. It is primarily characterised by atypical non-compliance and an extreme avoidance of everyday demands and requests. Although originally proposed

as a standalone diagnosis, there has since been a paucity of research into PDA to support this. The limited evidence available suggests that PDA is best understood as a set of traits occurring dimensionally within ASC (Duncan, Healy, Fiddler & Christie, 2011; Garralda, 2003; Green et al., 2018).

In addition, other traits reported in accounts of PDA include a lack of deference to authority, emotional lability associated with the need for control, and appearing comfortable in role-play and with pretence. Children fitting this profile reportedly display a 'surface' sociability but with an apparent lack of social identity, pride, or shame; represented by difficulties with peer interactions and a lack of social constraint (Newson et al., 2003). Specific demand avoidant traits can be categorised by attempts to subvert requests using distraction, social manipulation or behaviour intending to shock or upset (O'Nions, Christie, Gould, Viding & Happe, 2014). As such, children displaying high levels of PDA traits often present with extremely challenging behaviour. These difficulties significantly impact everyday life and are associated with some of the highest rates of exclusions from school and significant parental stress (Gore-Langton & Frederickson, 2015; Gore-Langton & Frederickson, 2018).

At a surface level, many of the traits associated with PDA presentations appear to overlap with those seen in common comorbid presentations of ASC; anxiety disorders, oppositional defiant disorder (ODD) and high callous-unemotional traits (Green et al., 2018; O'Nions, Viding, Greven, Ronald & Happe, 2014; Salazar et al., 2015). However, there is very little research into the underlying processes behind these behaviours in PDA. If we consider again the concept of equifinality, it may be too simplistic to suggest that these symptoms occur in ASC simply as a result of the same underlying mechanisms. Thus, given the well documented impact of the difficulties associated with PDA, and an increase in use of the term in UK health and education

settings (O'Nions et al., 2016), there is a clear rationale for further research into the profile. In particular, further understanding of the mechanisms behind PDA behaviours would contribute valuable insight into the clinical validity of the term and the relation to other presentations within ASC.

2.2.3 Theory of Mind (ToM)

Atypicality in cognitive processing is thought to underpin impairments in social interaction and behaviours observed in ASC (Chevallier, Kohls, Troiani, Brodkin & Schultz, 2012). Of particular relevance is the role of ToM. ToM is a socio-cognitive construct defined as the ability to attribute mental states to ourselves and others, and to use this to explain behaviour (Premack & Woodruff, 1987) based on the understanding that the minds of others differ to our own (Frith & Frith, 2005). This knowledge; that emotions, beliefs, and experiences inform behaviour, is thought to be key in developing an accurate understanding of human interaction and motive (Wellman, Cross & Watson, 2001).

ToM can be measured in a number of ways and is generally understood in terms of a composite score on tasks assessing different facets of the construct including: recognising and discriminating between emotions, understanding mental state terms, and having a knowledge of the pragmatic and metalinguistic aspects of language (Happe & Frith, 2014). ToM understanding is developmental, with different capabilities emerging at different stages in childhood. Early, cognitive facets of ToM understanding are thought to be evident from as young as nine months old in typically developing infants (Baron-Cohen, 1991), with more affective ToM capabilities continuing to emerge into adolescence (Vetter, Altgassen, Phillips, Mahy & Kliegel, 2013). There is an increasingly popular school of thought that ToM should only be

understood in terms of this distinction between 'cognitive' and 'affective' subcomponents (Shamay-Tsoory, Harari, Aharon-Peretz & Levkowitz, 2010).

It is widely agreed that children with ASC show impairments across all areas of ToM (Baron-Cohen, Wheelwright, Hill, Raste & Plumb, 2001), with some evidence to suggest that core ASC traits are predictive of ToM ability (Ronald, Viding, Happe & Plomin, 2007). The relationship between ToM and ASC traits is particularly salient to the understanding of impaired social interactions and communication difficulties observed in ASC (Baron-Cohen, Leslie & Frith, 1985), and perhaps less so for the repetitive and restrictive behaviours which complete the dyad of symptomology. Although it is generally agreed that children with ASC typically show impaired ToM, there is also significant variability in profiles (Happe, 1995). There is an argument to suggest that this variability is more indicative of difficulties initiating a spontaneous tracking of one's own and others mental states in ASC (Happe, 2003; Senju, 2011) as opposed to a 'lack' of ToM ability. Thus, the inference of mental states in ASC may be more deliberative, effortful, and less efficient resulting in errors and social faux pas.

2.2.4 ToM and PDA Traits

Newson et al. (2003) argued that many of the behavioural difficulties associated with PDA traits are likely to occur in relation to differences in socio-cognitive processes, specifically difficulties in recognising hierarchy and social identification/affiliation with peers (Happe & Frith, 2014). However, conceptually there is evidence to suggest that many specific PDA traits could be understood in terms of ToM.

Firstly, extreme demand avoidance is thought to be facilitated by a degree of sociability which allows for the use of socially controlling or manipulative strategies (Newson et al., 2003). To use strategic behaviour for personal gain is thought to rely

on a well-developed understanding of the minds of others, associated with better-developed ToM skills (Bosse, Memon & Treur, 2007; Sutton, Smith & Swettenham, 1999). This suggests that PDA traits may require better-developed ToM skills - in contrast to that which is observed in more typical presentations of ASC (Williams White, Keonig & Scahill, 2007). However, children with high levels of PDA traits also present with the same impairments in social behaviours that are observed in more typical presentations of ASC (O'Nions et al., 2014). Given that difficulties in these areas have been linked to ToM deficits (Hughes & Leekam, 2004; Imuta, Henry, Slaughter, Selcuk & Ruffman, 2016), this suggests impairments in areas of ToM (particularly with respect to internalising behaviour) may be associated with higher levels of PDA traits. This alludes to the possibility of a complex, multi-faceted role for ToM in understanding PDA traits.

Secondly, PDA is reportedly characterised by a positive response to humour, spontaneity and unpredictability – in contrast to the structured and predictable strategies traditionally employed to manage behaviour in ASC (Kunce & Mesibov, 1998). Appreciating humour is thought to relate positively to ToM (Uekermann, Channon, Winkel, Schlebusch & Daum, 2007), specifically as it relies on an understanding of pragmatics (the use of contextually appropriate language in interactions) (Tager-Flusberg, 2000); an important facet of ToM (Hutchins, Prelock & Bonazinga, 2011). Managing spontaneity and unpredictability is thought to require cognitive and social flexibility, abilities which are linked to ToM in typically developing children (Farrant, Fletcher & Maybert, 2014; Jacques & Zelazo, 2005). Social flexibility refers to deficits in social interaction and a rigid adherence to routine; features observed in typical ASC presentations (Chowdhury et al., 2016). As children with high PDA traits have been observed to respond better to spontaneity and

unpredictability, this suggests the profile may be associated with increased social flexibility than is more typically observed in ASC. This implies that this trait in PDA may be associated with better ToM.

Finally, being comfortable in role-play and fantasy was described in some accounts of PDA (Newson et al., 2003). This is in direct contrast to the absent or delayed pretend play associated with more typical ASC presentations (Frith, Morton & Leslie, 1991; Rutherford & Rogers, 2003). Previous research has argued for the importance of ToM in role-play and fantasy, suggesting that ToM acts as a cognitive precursor to pretence and as such, must have developed to some extent to allow the 'decoupling' of primary representations from pretend (Leslie, 1987). This suggests an engagement in pretence and fantasy may require ToM capability. However, there is also a school of thought that children with high PDA trait presentations can become preoccupied with pretending and fantasy, and struggle to separate this from reality (O'Nions et al., 2014), suggesting the relationship may be more complex than simply depending on a well-developed ToM understanding.

Thus, there are reasons to hypothesise that there may be a link between PDA traits and ToM in children with ASC, with some PDA traits associated with better ToM than is typically observed in ASC.

2.2.5 Rationale

The increased use of the PDA label as a clinical descriptor has sparked a need for empirical and systematic investigation of the epidemiology, diagnostic boundaries and treatment of the presentation (Gillberg, 2014). Although there are a small number of studies investigating PDA traits in ASC (e.g. O'Nions et al., 2014), there is a relative lack of research exploring the socio-cognitive processes behind these traits.

ToM is a socio-cognitive capacity that has been implicated in understanding behavioural difficulties associated with typical presentations of ASC. Many PDA traits seem to contrast with core ASC traits, and conceptually, there is evidence to hypothesise a link between PDA specific traits and ToM. As such, further investigation of the association between ToM and specific PDA traits, and between ToM and core ASC traits would help to provide knowledge of the underlying mechanisms and processes behind these behaviours. This would contribute to an understanding of the underlying routes to the typical and atypical behaviours observed in ASC, and would inform the clinical utility of PDA as a descriptive term.

2.2.6 Aims

The over-arching aim of this study was to contribute to an increased understanding of the association between ToM and traits and behaviours relevant to PDA and ASC. As such, the aims of the study were as follows:

1. To explore differences in ToM between control participants and participants with ASC and varying levels of PDA traits.

2. To investigate the relationship between ToM and (1) traits and behaviours relevant to PDA, and (2) core ASC traits within a group who have received an ASC diagnosis.

It was expected that control participants would have better ToM abilities than ASC participants. In addition, it was expected there would be an association between ToM and behaviour and traits relevant to PDA, specifically, that there would be a positive relationship between engaging in pretence and fantasy and ToM, and between ToM and social flexibility. In addition, it was hypothesised that socially manipulative behaviour and demand specific non-compliance would be associated with ToM skills in children with ASC. Finally, due to the well-documented links between ToM and

ASC, it was predicted that there would be a negative relationship between core ASC traits and ToM in the ASC sample.

Given the considerable interest in high PDA trait presentations within ASC in UK clinical settings, the final aim for this study was:

3. To investigate differences between high and low PDA presentations within an ASC sample.

2.3 Method

2.3.1 Design

The study employed a non-experimental, cross-sectional design with continuous and between-group analyses. It formed part of a joint thesis for the doctorate in clinical psychology at University College London. The other submission focused on an investigation of the relationship between PDA traits in ASC and executive functioning, and was carried out by Anna Goodson. A breakdown of the individual trainee contributions to joint working can be found in Appendix A.

2.3.2 Ethical Approval

Ethical approval was granted for this study by the University College London Research Ethics Committee (REC Reference: 10193/001, Appendix B). Participants volunteered to take part and informed consent was obtained from all participants before any data was collected. Participant information sheets and consent forms can be found in Appendices C and D.

2.3.3 Participants

Power calculations were conducted to infer an appropriate sample size for this study. Based on the assumption of attaining a medium to large effect size for between-

group analyses (O'Nions et al., 2014; Cohen, 1992), a sample size of larger than 60 was deemed sufficient to detect effects with a specified alpha of .05 and power of 80%.

Based on this, we aimed to recruit parents of 60 children with ASC and 30 non-ASC controls. As such, the participants for this study were the parents of 91 children aged between 6-11 years old (M=8 years, 9 months; SD= 1 year, 7 months). Sixty-one participants reported their child had a diagnosis of ASC and 30 participants reported their child met criteria for inclusion in a non-ASC control group. Full details of participant characteristics are described in Table 2.1.

2.3.4 Procedure

2.3.4.1 Recruitment. We aimed to recruit parents of children aged 6-11 years old, who could either be classified as non-ASC controls or who reported a diagnosis of ASC, with or without PDA traits. To ensure a broad range of PDA symptomology in the ASC sample, recruitment was monitored so that approximately half of the participants reported a score above the clinical threshold on a measure of PDA traits.

Participants were recruited via opportunity sampling and snowballing recruitment techniques. The study was promoted online using a variety of non-NHS platforms, primarily advertised by the PDA Society, the National Autistic Society, Mumsnet, and various parenting groups on social media sites. The advertisement poster for the study can be found in Appendix E.

Parents were screened for eligibility based on initial inclusion and exclusion criteria. Parents were eligible to participate on the condition that they were (1) over 18 years of age (2) English speaking and (3) had a child aged 6-11 years old. Parents of children diagnosed with a moderate to severe learning disability were excluded from the study. In addition, parents' responses to an initial set of questionnaires made it

possible to screen participants against criteria for inclusion in an ASC or non-ASC control study group, defined as follows:

- Non-ASC control = No previous diagnosis of ASC indicated by:

    1. Parent-report of no diagnosis

    2. A score below the cut-off of 15 on the Childhood Autism Spectrum Test (CAST; Scott, Baron-Cohen, Bolton & Brayne, 2002)

    3. A score below the cut-off of 50 on the Extreme Demand Avoidance Questionnaire (EDA-Q; O'Nions et al., 2014).

    4. A score below the cut-off of 16 on the Overall Difficulties subscale of the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997)

- ASC = Parent-report of a previous diagnosis of ASC from a professional.

The parents of 208 children were screened for eligibility based on these criteria. The parents of 98 children did not meet criteria for inclusion in either study group and as such, were not eligible to participate. Therefore, 110 parents were recruited on a first come, first served basis. Ninety-one parents completed all measures and formed the participant group for this study.

2.3.4.2 Study procedure. Participation was conducted using a two-phase approach and was entirely online. Data were collected via Qualtrics and through a secure internet site specifically for ToM reporting. Parents initially registered their interest in the study by emailing one of the research team or by providing an email address via a secure Qualtrics link. Interested parents were allocated a unique participant number and sent a password-protected link to an online participant information sheet and consent form. A template for emails to participants is shown in Appendix F.

Once informed consent had been indicated, parents were sent a secure link to Phase 1 of the study. They were asked to respond to a demographic questionnaire, (shown in Appendix G) and to complete a set of questionnaires measuring core ASC traits, PDA traits, and emotional and behavioural difficulties. As described previously, responses to questionnaires at Phase 1 determined whether participants were eligible to complete Phase 2 of the study. Eligible participants were then sent an email with a secure link to access Phase 2 of the study, comprising two further questionnaires measuring ASC and PDA behaviours, and ToM. The data from non-eligible and non-complete participants was not included in the analysis.

2.3.4.3 Measures. The following constructs were assessed at Phase 1 using standardised measures.

Core ASC traits. Core ASC traits were assessed using the CAST (Scott et al., 2002; Appendix H). The CAST is a 37-item, parent-report questionnaire to screen for ASC traits. Items are presented as a 'yes' or 'no' question (e.g. can s/he hold a two way conversation?) and scored either 0 or 1. It has been shown to have good reliability and validity, with a cut-off score of 15 or above indicating risk of ASC with 100% sensitivity and 97% specificity (Williams et al., 2005). The CAST has three distinct subscales assessing specific ASC traits; Social Difficulties, Communication Problems and RRBI's (Ronald, Happe, Price, Baron-Cohen & Plomin, 2006).

Emotional and behavioural difficulties. Emotional and behavioural difficulties were measured using the SDQ (Goodman, 1997; Appendix I). The SDQ is a parent-report questionnaire, valid for use in the framework of multi-dimensional behavioural assessment (Becker, Woerner, Hasselhorn, Banaschewski & Rothenberger, 2004; Goodman, 2001) in both typically developing and ASC populations (Simonoff et al., 2012). The SDQ comprises 25 items phrased as

statements (e.g. 'my child has many fears and is easily scared'), which are scored on a Likert scale (0=Not True to 2=Certainly True). It reports an Overall Difficulties score, with a threshold of over 16 indicating the presence of clinically relevant emotional and behavioural difficulties. In addition, there are five distinct subscales used to report difficulties in different areas (Emotional, Conduct, Hyperactivity, Peer Relationships and Prosocial Behaviour subscales), and a further two subscales suggested for use in general populations where there is likely to be less clinical need (Internalising and Externalising).

PDA traits. PDA traits were assessed using the EDA-Q (O'Nions et al., 2014; Appendix J); a 26-item parent-report measure of extreme demand avoidance and other PDA behavioural features described by Newson et al. (2003). Items are presented as a statement (e.g. 'uses outrageous or shocking behaviour to get out of doing something) and scored on a response scale from 0=Not True to 3=Very True. It has a good degree of sensitivity (80%) and specificity (85%) to identify children at an elevated risk of having a profile consistent with descriptions of PDA. The measure has a high level of internal consistency and has been partially validated.

Further to the total scale score, two additional subscales were generated from the EDA-Q items for use in this study: 'Social Manipulation' (six items) and 'Pretence and Fantasy' (four items) (Appendix M). These subscales were derived based on item content and agreed separately by three researchers. Both had a good degree of internal consistency (α=.902 and α=.837 respectively).

The following measures were completed at Phase 2:

ASC/PDA behaviours. ASC and PDA behavioural difficulties in everyday settings were assessed using the Home Situations Questionnaire – ASD (HSQ-ASD; Barkley & Edelbrock, 1987; Chowdhury et al., 2015; Appendix K). The HSQ-ASD

reports a total score and can be analysed using two subscales, Social Inflexibility and Demand Specific Non-Compliance. It measures non-compliance with instructions, commands, or rules in everyday situations that may be problematic for children with ASC and PDA traits (e.g. routine requests, sensory challenges, and social situations with a high degree of uncertainty). The HSQ-ASD has 24 constituent items pertaining to a child's difficulties with day to day activities (e.g. responses to household rules). Items are responded to on a yes or no scale. If the response is 'yes', parents are asked to indicate a degree of severity from 1=Mild to 9=Severe. It has been shown to be a good identifier of individual variability.

ToM. ToM was assessed using the Theory of Mind Inventory 2 (ToMI-2; Lerner, Hutchins & Prelock, 2011; Appendix L). The ToMI-2 is a well-validated and reliable measure that seeks to assess a wide range of ToM competencies based on informant-report (Greenslade & Coggins, 2016; Hutchins et al., 2012). Informant-report is deemed to be a valid and reliable measure of ToM (e.g. Tahiroglu et al., 2014). Participants completed the ToMI-2 online via a purpose designed, secure platform. Each item is presented as a statement (e.g., "My child understands whether someone hurts another on purpose or by accident"), which is scored on a sliding scale ranging from 'Definitely' to 'Definitely Not'.

The ToMI-2 can be analysed using three empirically derived subscales: Early, Basic and Advanced ToM. Early ToM taps skills in reading affect, sharing attention and recognising intent. These are usually evident in a typically developing population from infancy and toddlerhood. Basic ToM taps false-belief and meta-representation understandings, evidence of which is observed in typically developing pre-schoolers from around the age of three. Advanced ToM measures complex recursion, metapragmatic, and metalinguistic skills thought to emerge in a typically developing

population from the age of six. Although these subscales refer to three developmental levels, they are not considered discrete or inflexible stages, and skills from each can emerge at different stages.

2.3.5 Data Analysis

As discussed, the data were collated and grouped according to ASC diagnosis. Tests of distribution were carried out to ensure the appropriateness of parametric analysis. The distributions for most variables did not significantly deviate from normality, reporting skew and kurtosis figures inside the $z=\pm3.29$ level recommended for this sample size (Field & Miles, 2009; Kim, 2013).

To assess the first aim, analyses were conducted using data from the ToMI-2. As the scores for the ASC and non-ASC control groups combined reported bimodal distributions and did not respond to transformation, non-parametric analysis was conducted to compare ToM between groups. The standard scores on the Composite index of the ToMI-2 were used for analysis, alongside the standard scores on the empirically derived subscales (Early, Basic and Advanced ToM).

To assess the second aim, separate correlation and regression analysis were performed to investigate the association between ToM and PDA traits, and between ToM and core ASC traits in the ASC sample alone. To quantify ToM, the Composite scale, and Early, Basic, and Advanced subscales on the ToMI-2 were used in the analysis. The Early and Basic subscales showed a significant level of positive skew for children with ASC. As such, they were transformed using a square root and logarithm transformation respectively. PDA traits and non-compliance behaviours were measured using the EDA-Q (total score, Social Manipulation and Pretence and Fantasy subscale scores) and the HSQ-ASD (total score, Social Inflexibility and Demand Specific subscale scores). Core ASC traits were measured using the CAST

(total score, Social Difficulties, Communication Problems, and RRBI subscale scores). The Communication Problems subscale showed a significant level of negative skew for children with ASC and was transformed using an inverse, square root transformation and re-reflected (Tabachnick & Fidell, 2007).

To explore our final aim, between-group analyses were conducted for individuals with ASC with low and high levels of PDA traits. The EDA-Q was used to categorise the ASC sample into two groups; high PDA traits (ASC+PDA) and low PDA traits (ASC-PDA). As the EDA-Q is not considered a diagnostic tool, this was done using a median-split analysis (e.g. Viding et al., 2012) with reported scores ≤ 53 classified as low PDA traits and reported scores >53 classed as high PDA traits. Descriptive statistics, parametric and non-parametric analyses were used to explore differences in emotional and behavioural difficulties, core ASC traits and ASC/PDA related behavioural difficulties between groups.

2.4 Results

2.4.1 Sample Characteristics

Table 2.1 reports demographic and behavioural characteristics for the sample. Differences in presentation between children with ASC and non-ASC controls were investigated.

The ASC and non-ASC groups were matched for age. However, there was a significantly increased ratio of boys to girls in the ASC group compared to non-ASC controls. Chi-squared tests of independence were conducted to compare the likelihood of being in mainstream education and having additional diagnoses between ASC and non-ASC children. As the assumptions for chi-squared were violated in that one or more cells had an expected value of 0, the Fisher's Exact Test statistic was used to

infer significance. Non-ASC children were significantly more likely to be in mainstream education (100%) compared to children with ASC (54.1%). Children in the ASC group were significantly more likely to have a comorbid diagnosis (32.7%) compared to children in the non-ASC group (6.7%).

Differences in emotional and behavioural difficulties were explored between groups using independent sample t-tests. Although scores on the SDQ were used to infer eligibility for the non-ASC control group, this was not the case for the ASC group. As such, between-group analyses were deemed appropriate to demonstrate differences between children with ASC and controls with no significant emotional or behavioural difficulties. Overall, children with ASC had significantly higher levels of emotional and behavioural difficulties compared to the non-ASC controls ($t(89)=-15.66$, $p<.001$, $d=3.64$). In addition, children with ASC showed significantly more difficulty on the Emotional, Conduct, Hyperactivity, Peer Relationships and Prosocial Behaviour subscales. There were also differences in the expression of these difficulties, with children with ASC demonstrating significantly more Internalising and Externalising difficulties than non-ASC controls ($t(89)=-14.43$, $p<.001$, $d=3.46$; $t(89)=-9.82$, $p<.001$, $d=2.24$ respectively).

Table 2.1. Demographic and clinical characteristics of participants

Measure	Group				Analysis		
	1. Non-ASC Controls (N=30)		2. ASC (N=61)				
	N	%	N	%	χ^2	p value	Φ
Gender					4.64	p<.05	-.23
Male	11	36.7	37	60.6			
Female	19	63.3	24	39.4			
Education					19.89	p<.001	.47
Mainstream Education	30	100	33	54.1			
Home School			11	18.0			
Specialised Unit / School			10	16.4			
Not in Education			2	3.3			
Other			5	8.2			
Additional Diagnoses					4.85	p<.05	.23
None	28	93.3	41	67.2			
ADHD			6	9.8			
Mild LD			4	6.6			
Learning Difficulties	2	6.7	3	4.9			
Other			7	11.5			
	M	SD	M	SD	t	p value	d
Age (months)	101.77	18.83	109.72	18.94		n.s	
SDQ Total	7.20	3.85	24.00	5.26	-15.66	1 < 2*	3.64
Emotional Problems	2.27	1.84	7.00	2.29	-9.87	1 < 2*	2.28
Conduct Problems	1.20	1.40	4.54	1.91	-8.51	1 < 2*	2.00
Hyperactivity	3.07	2.29	7.03	2.45	-7.42	1 < 2*	1.67
Peer Problems	.67	1.03	5.43	2.00	-12.22	1 < 2*	2.99
Prosocial Behaviours	8.53	2.33	3.70	2.33	10.45	1 > 2*	2.07
Internalising Problems	2.93	2.05	12.43	3.30	-14.43	1 < 2*	3.46
Externalising Problems	4.27	3.06	11.57	3.46	-9.82	1 < 2*	2.24

* Significant at *p*<.001 level

2.4.2 Aim 1 – Group Differences in ToM in ASC

To assess the first aim, Mann-Whitney analyses were conducted to assess ToM differences between the ASC group (n=61) and non-ASC controls (n=30).

Composite ToM scores were reported to be significantly better for children in the non-ASC group compared to the ASC group (*U*=89, *p*<.001, *r*=.73). In addition, significantly better ToM scores were reported for children in the non-ASC group on the Early, Basic, and Advanced subscales (Early *U*=204.5, *p*<.001, *r*=.63; Basic *U*=236, *p*<.001, *r*=.60; Advanced *U*=35.5, *p*<.001, *r*=.78). Due to the gender discrepancy between the ASC and non-ASC groups, these analyses were re-conducted by gender. The findings were consistent with the overall outcome and did not significantly differ by gender.

The distributions and range of ToM scores for the ASC and non-ASC groups are depicted in Figure 2.1. Significant outliers were highlighted in the ASC group on the Basic and Advanced subscales. However, as these scores were feasible (e.g. ToM capacity varies across ASC), they were not removed from the analyses.
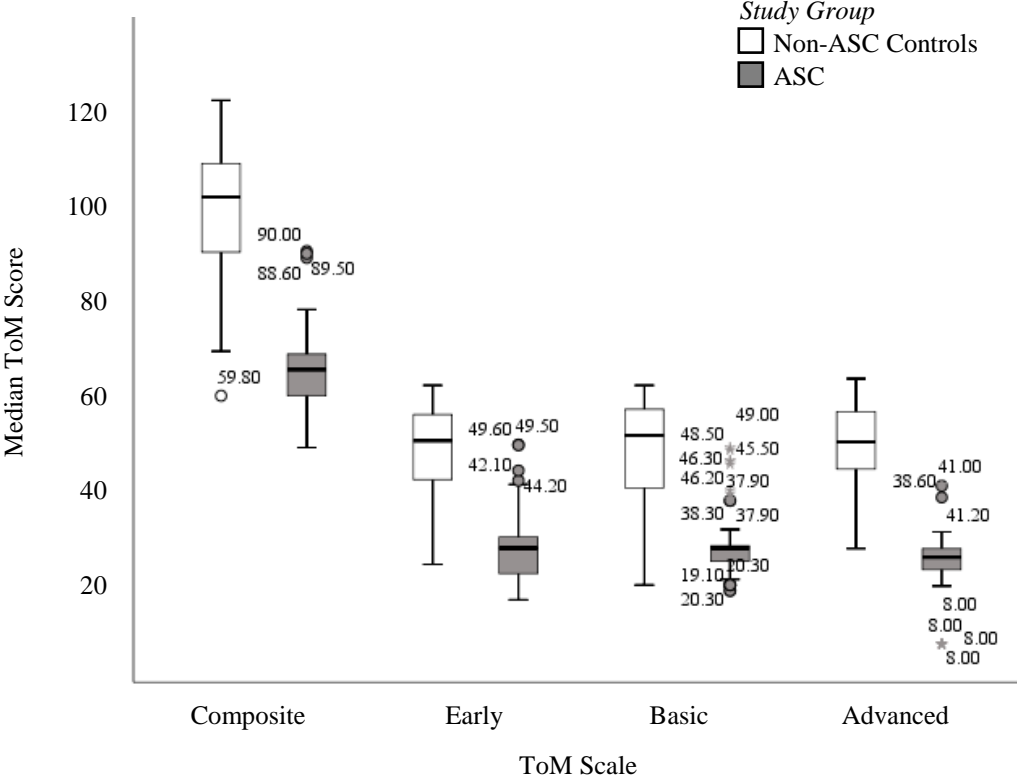
Figure 2.1 Box-plot showing the distribution of ToM scores for the ASC and non-ASC control groups.

In addition, there appeared to be limited variation in the percentile data for the ASC sample on the ToMI-2, indicative of a floor effect. 85.2% of respondents indicated scores at the 1st percentile for Composite ToM, 86.9% for Early ToM, 72.1% for Basic ToM and 83.6% for Advanced ToM. This was not the case for the non-ASC sample, where scores were largely in line with norms reported for typically developing children (Hutchins et al., 2012).

2.4.3 Aim 2 – ToM and (1) PDA traits and (2) core ASC traits

Separate correlation and regression analyses were conducted in accordance with our second aim – to investigate the relationship between ToM and (1) PDA traits and (2) core ASC traits in the ASC sample.

2.4.3.1 ToM and PDA traits. Analysis within the ASC group was conducted to investigate the relationship between ToM and PDA traits. Correlation analyses were used to identify associations between ToM and PDA traits, as shown in Table 2.2. Age and gender were included in the matrix to assess for significance.

As the ToMI-2 reported standard scores, age was accounted for and as such, we did not expect there to be any significant correlation between age and the ToM variables. However, there was a nominally significant correlation between age and the Basic subscale ($r=.270$, $p<.05$), suggesting better Basic ToM abilities were reported for older children with ASC in comparison to the other ToM scales. There was no association between age and any measure of PDA traits, with the exception of Pretence and Fantasy, which returned a significant positive correlation ($r=.315$, $p<.05$), suggesting that older children show more behaviours associated with pretence and fantasy. Although gender is a binary variable, correlations between gender and ToM and PDA traits were deemed appropriate based on previous studies employing the same methodology (e.g. Choe, Lane, Grabell & Olson., 2013). There was no association between gender and PDA traits. There was a nominally significant correlation between gender and the Basic ToM subscale ($r=.324$, $p<.05$). As none of the correlations between age or gender and ToM or PDA traits would survive correction for multiple comparison, age and gender were not included as covariates in the correlation analyses.

Overall, the data did not suggest a significant relationship between scores on the ToMI-2 and measures of PDA traits. There was some evidence suggestive of a

weak negative correlation between Basic and Advanced ToM, and the Social Inflexibility and Demand Specific subscales of the HSQ-ASD. However, as multiple correlations were conducted, the alpha level at which significance is inferred needed to be interpreted with caution. Although conservative, Bonferroni correction was deemed the most appropriate method to control for multiple correlation comparisons (Curtin & Schulz, 1998). As no correlations remained significant following Bonferroni correction ($p<.002$) further regression analysis was not deemed appropriate.

2.4.3.2 ToM and core ASC traits. Correlation analysis was conducted within the ASC group to investigate the relationship between ToM and core ASC traits, shown in Table 2.3.

Again, age and gender were included to assess for any confounding influence on the data. There was a significant relationship between higher scores on total scale CAST and gender ($r=-.320$, $p<.05$), and RRBI scores and gender ($r=-.414$, $p<.001$), both suggesting that lower scores were reported for females than males. As the correlation between gender and total scale CAST scores did not survive correction for multiple comparison, gender was not included as a covariate in the correlation analyses.

Correlation analysis revealed that better ToM scores were associated with lower levels of ASC traits. As multiple correlations were conducted, significance was interpreted at an alpha level of $p<.003$, as specified by Bonferroni correction. There were significant negative correlations between scores on the Composite, Basic and Advanced ToM scales and the total score on the CAST ($r=-.383$, $p<.001$; $r=-.414$, $p<.001$; $r=-.407$, $p<.001$ respectively). At the subscale level, there were significant negative correlations between the Social Difficulties subscale and Basic ToM ($r=-.370$, $p<.001$), and the Communication Problems subscale and (1) Composite ToM

(r=-.436, p<.001) and (2) Advanced ToM (r=-.409, p<.001). As the relationship between the RRBI subscale and ToM was not significant, the role of gender was not assessed further.

Linear regression analyses were carried out to assess whether reports of core ASC traits were predicted by ToM scores. As the relationship between gender and total scale CAST scores was approaching significance even after Bonferroni correction, hierarchical linear regressions including gender were conducted for this scale. The data were assessed to confirm they met the assumptions for regression outlined by Field (2009). As the Durbin-Watson values were close to 2, independent errors between residuals were assumed. Assumptions of no multicollinearity were met, as assessed by VIF and tolerance statistics. In addition, a visual inspection of the plotted residuals suggested the data did not violate the assumption of homoscedasticity.

As shown in Table 2.4, Composite ToM was found to be a significant predictor of core ASC traits as measured by the total score on the CAST ($F(1, 59)$=10.13, p<.01). This means that every unit increase in Composite ToM was associated with a .21 unit decrease in total CAST score, with ToM accounting for 14.7% of the variance in total CAST scores. Composite ToM and gender combined predicted overall CAST scores to a significant level ($F(2, 58)$=8.26, p<.001), accounting for 22.2% of the variance in the total score. Total CAST scores were also independently predicted by Basic ToM skills ($F(1, 59)$=12.22, p<.001; 17.2% of the variance) and Advanced ToM skills ($F(1, 59)$=11.74; p<.001; 16.6% of the variance).

In addition, scores on the Social Difficulties subscale were independently predicted by Basic ToM scores ($F(1, 59)$=9.34, p<.01; 13.7% of the variance). Transformed scores on the Communication Problems subscale were independently

predicted by scores on the Composite ToM index ($F(1, 59)=14.26$, $p<.001$; 19.5% of the variance) and Advanced ToM skills ($F(1, 59)=10.38$, $p<.01$; 15% of the variance).

Table 2.2. Correlation matrix for ToM and PDA traits

	ToM				Pathological Demand Avoidance Traits					
	1	2	3	4	5	6	7	8	9	10
Age	.122	.123	.270*	.016	-.080	.082	.315*	-.017	-.074	.040
Gender[1]	.124	-.058	.324*	.156	-.106	-.088	.032	-.039	-.056	-.019
ToM										
1. Composite Score		.385**	.366**	.776**	-.184	-.129	-.243	-.120	-.104	-.106
2. Early ToM			.395**	.243	-.194	-.061	-.200	-.103	-.077	-.115
3. Basic ToM				.218	-.175	-.085	-.091	-.292*	-.321*	-.232
4. Advanced ToM					-.185	-.148	-.063	-.250	-.174	-.265*
PDA Traits										
5. EDA-Q Total						.874**	.691**	.657**	.649**	.571**
6. Social Manipulation							.539**	.592**	.558**	.532**
7. Pretence and Fantasy								.223	.275*	.146
8. HSQ-ASD Total									.929**	.944**
9. Social Inflexibility										.756**
10. Demand Specific										

* Significant at the $p<.05$ level
** Significant at the $p<.001$ level
[1] Male = 0, Female = 1

Table 2.3. Correlation matrix for ToM and core ASC Traits

	ToM				Core ASC Traits			
	1	2	3	4	5	6	7	8
Age	.122	.123	.270*	.016	-.089	-.061	-.133	-.008
Gender[1]	.124	-.058	.324*	.156	-.320*	.233	-.182	-.414**
ToM								
1. Composite Score		.385**	.366*	.776**	-.383**	-.225	-.436**	-.278*
2. Early ToM			.395**	.243	-.278*	-.291*	-.170	-.138
3. Basic ToM				.218	-.414**	-.370**	-.299*	-.298*
4. Advanced ToM					-.407**	-.304*	-.409**	-.298*
Core ASC Traits								
5. CAST Total						.853**	.842**	.699**
6. Social Difficulties							.545**	.378**
7. Communication Problems								-.505**
8. RRBIs								

* Significant at the $p<.05$ level
** Significant at the $p<.001$ level
[1] Male = 0, Female = 1

Table 2.4. Regression analysis for ToM and gender predicting core ASC traits

		Unstandardised Coefficients		Standardised Coefficients					
	Variable	B	SE	B	p value	R^2	R^2 change	F	p value
Total Score CAST									
Model 1	Step 1					.147	.147	10.13	<.01
	Composite ToM	-.21	.07	-.38	<.01				
	Step 2					.222	.075	8.26	<.001
	Composite ToM	-.19	.06	-.35	<.01				
	Gender	-2.80	1.18	-.28	<.05				
Model 2	Step 1					.172	.172	12.22	<.001
	Basic ToM	-20.98*	6.00	-.41	<.001				
	Step 2					.210	.038	7.71	<.001
	Basic ToM	-17.58*	6.25	-.35	<.01				
	Gender	-2.09	1.25	-.21	n.s				
Model 3	Step 1					.166	.166	11.74	<.001
	Advanced ToM	-.28	.08	-.41	<.001				
	Step 2					.233	.067	8.81	<.001
	Advanced ToM	-.25	.08	-.37	<.01				
	Gender	-2.66	1.18	-.26	<.05				
CAST Subscale scores									
Social Difficulties									
	Basic ToM	-9.65*	3.16	-.37	<.01	.137		9.34	<.01
Communication Problems**									
	Composite ToM	.10	.03	.44	<.001	.195		14.26	<.001
	Advanced ToM	.11	.03	.39	<.01	.150		10.38	<.01

* Non-transformed predictor variable B statistic
** Non-transformed outcome variable B statistic

Table 2.5 Demographic and clinical characteristics of ASC-PDA and ASC+PDA

Measure	Group						Analysis		
	1. ASC-PDA (N=31)			2. ASC+PDA (N=30)					
	N	%		N	%		χ^2	p value	Φ
Gender							.89	n.s	.12
Male	17	54.8		20	66.7				
Female	14	45.2		10	33.3				
Education									
Mainstream Education	20	64.5		13	43.3		2.76	n.s	.21
Home School	4	12.9		7	23.3				
Specialised Unit / School	5	15.1		5	16.6				
Not in Education				2	6.7				
Other	2	6.5		3	10.0				
Additional Diagnoses									
None	23	74.2		18	60		1.39	n.s	.15
ADHD	1	3.2		5	16.7				
Mild LD	3	9.7		1	3.3				
Learning Difficulties	1	3.2		2	6.7				
Other	3	9.7		4	13.3				
	M	SD	Range	M	SD	Range	t	p value	d
Age (months)	111.29	18.80	74 – 142	108.10	19.27	74 – 143		n.s	
SDQ Total	20.97	4.38	13 – 29	27.13	4.51	20 – 37	-5.70	1 < 2**	1.39
Emotional Problems	6.48	2.52	2 – 10	7.53	1.96	2 – 10		n.s	.46
Conduct Problems	3.42	1.50	0 – 6	5.70	1.58	3 – 10	-5.78	1 < 2**	1.48
Hyperactivity	6.23	2.58	1 – 10	7.87	2.03	3 – 10	-2.76	1 < 2*	.71
Peer Problems	4.84	1.99	1 – 9	6.03	1.87	2 – 9	-2.42	1 < 2*	.62
Prosocial Behaviours	4.10	2.24	1 – 10	3.30	2.38	0 – 8		n.s	.35
Internalising Problems	11.32	3.10	6 – 18	13.57	3.15	4 – 19	-2.80	1 < 2*	.72
Externalising Problems	9.65	3.03	5 – 15	13.57	2.69	7 – 18	-5.34	1 < 2**	1.37

Measure	Group						Analysis		
	1. ASC-PDA (N=31)			2. ASC+PDA (N=30)					
	M	SD	Range	M	SD	Range	t	p value	d
CAST Total	18.94	5.48	6 – 29	22.30	3.83	13 – 29	-2.77	1 < 2*	.71
Social Difficulties	6.42	2.72	2 – 12	7.07	2.42	3 – 12		n.s	.25
Communication Problems[1]	8.00	2.26	1 – 11	10.00	1.11	7 – 11	-4.76	1 < 2**	1.22
RRBIs	5.16	1.53	2 – 7	5.70	1.42	3 – 7		n.s	.37
HSQ-ASD	4.39	1.90	.75 – 8.25	6.39	2.00	4.21 – 8.42	-4.78	1 < 2**	1.03
Social Inflexibility	4.51	1.98	1.00 – 8.33	6.38	1.17	4.64 – 8.58	-4.42	1 < 2**	1.15
Demand Specific	4.27	2.11	.50 – 8.17	6.34	1.56	3.67 – 9.00	-4.25	1 < 2**	1.12
Composite ToM	65.81	9.53	49 – 90	64.45	9.10	49 – 88.6		n.s	.15
Early ToM[1]	28.00	7.33	17.2 – 49.5	29.00	6.50	17.2 – 49.6		n.s	.14
Basic ToM[1]	28.00	8.28	20.3 – 49	28.00	5.85	19.1 – 45.5		n.s	.22
Advanced ToM	25.64	7.39	8 – 41	23.84	7.27	8 – 38.6		n.s	.34

* Significant at $p<.05$ level

** Significant at $p<.001$ level

[1] Un-transformed median and range reported as measures of central tendency, t-tests on transformed scales

2.4.4 Aim 3 - Clinical Description of PDA Traits in ASC

Between-group analysis was conducted to compare children with diagnoses of ASC and low PDA traits (ASC-PDA) to those with high PDA traits (ASC+PDA), as shown in Table 2.5.

2.4.4.1 Education. ASC+PDA were more likely to require specialist educational provision than ASC-PDA, with only 43.3% accessing mainstream education, compared to 64.5%. Although this finding did not reach statistical significance (χ^2=2.76, p=.10 FET) the figures are likely to represent clinically significant differences in the level of educational need.

2.4.4.2 Additional diagnoses. There were no significant differences in the overall likelihood of having an additional diagnosis in the ASC+PDA group compared to the ASC-PDA group.

2.4.4.3 Emotional and behavioural difficulties. Significantly greater Overall emotional and behavioural difficulties were reported for ASC+PDA than for ASC-PDA (t(59)=-5.70, p<.001, d=1.39). At subscale level, significantly greater difficulties with Conduct, Hyperactivity and Peer Relationships were reported for ASC+PDA than ASC-PDA (t(59)=-5.78, p<.001, d=1.48; t(59)=-2.76, p<.06, d=.71; t(59)=-2.42, p<.05, d=.62 respectively). Additionally, significantly greater difficulty with Internalising and Externalising behaviours were reported for ASC+PDA compared to ASC-PDA (t(59)=-2.80, p<.05, d=.72; t(59)=-5.34, p<.001, d=1.37 respectively). There were no significant differences between ASC+PDA and ASC-PDA on reports of Emotional difficulties and Prosocial Behaviours.

2.4.4.4 Core ASC traits and PDA behaviours. Differences in core ASC traits and ASC specific behaviours were assessed using the total and subscale scores on the CAST and the HSQ-ASD. Overall, parents reported significantly higher total scale

scores on the CAST for ASC+PDA compared to ASC-PDA (t(59)=-2.77, p<.05, d=.71). Significantly higher scores were reported on the Communication Problems subscale for children with ASC+PDA compared to ASC-PDA (t(59)=4.81, p<.001, d=1.22). There were no significant differences between scores for ASC+PDA and ASC-PDA on the Social Problems and RRBI subscales.

Parents reported significantly higher levels of ASC and PDA related behavioural difficulties on the total scale score of the HSQ-ASD for ASC+PDA compared to ASC-PDA (t(57)=-4.78, p<.001, d=1.03). Significantly higher scores on the Social Inflexibility subscale (t(58)=-4.42, p<.001, d=1.15) and the Demand-Specific subscale (t(57)=-4.25, p<.001, d=1.12) were reported for ASC+PDA compared to ASC-PDA.

2.4.4.5 ToM. Reported ToM scores did not significantly differ between children in the ASC-PDA and ASC+PDA groups. In addition, the distribution and range of ToM scores did not differ significantly between groups.

2.5 Discussion

This study aimed to provide further understanding of PDA traits in ASC, and specifically to investigate the relationship between ToM and traits and behaviours relevant to PDA and ASC. Parent-report measures were analysed using correlation and between-group analyses. The results of this study will be considered with reference to the wider literature and implications for the conceptualisation of PDA traits in ASC. The limitations arising from the design and methodology will be discussed and conclusions on the study as a whole will be drawn.

2.5.1 ToM, PDA and ASC Traits

As expected, parents reported significantly better-developed ToM across all scales for children without ASC compared to those with an ASC diagnosis.

Overall, there was no significant association between parent-report ToM scores and measures of PDA traits. Although there was a nominally significant relationship between better Basic and Advanced ToM and lower levels of PDA behaviours reported on the HSQ-ASD, this finding did not meet the threshold for significance following correction for multiple comparisons. In contrast to our specific predictions, scores on the Pretence and Fantasy and Social Inflexibility subscales did not relate to better ToM capacity. In addition, there was no relationship found between scores on the Social Manipulation and Demand Specific Non-Compliance subscales and ToM.

In children with ASC, better parent-reported ToM was significantly associated with lower levels of core ASC traits, and specifically with lower scores on the Social Difficulties and Communication Problems subscales of the CAST. Different subscales of ToM predicted overall levels of core ASC traits, Social Difficulties and Communication Problems to varying degrees of significance, accounting for between 13-20% of the variance in scores. The relationship between ToM and RRBIs did not survive correction for multiple comparisons. In addition, Early ToM abilities (e.g. reading affect, sharing attention, and inferring intent) did not significantly relate to core ASC traits.

2.5.2 Interpretation of Findings

As predicted, ToM scores were negatively related to overall rates of core ASC traits, and to social and communication impairments in ASC. However, there was no significant relationship found between PDA traits and ToM. Theoretical suggestions and potential explanations for these findings will be considered further.

Although ToM scores significantly predicted measures of core ASC traits, there was a large amount of residual variance in CAST scores which was not explained by this study. The most parsimonious explanation for this finding is that there are simply other mechanisms underlying both core ASC and PDA traits and behaviours. Executive functioning and central coherence have both been associated with ASC traits (Happe & Frith, 2006; Kenworthy, Black, Harrison, Della Rosa & Wallace, 2008; Pellicano, 2010) and conceptually, could be implicated in explanations of PDA features as observed by Newson et al. (2003). For example, the behavioural disinhibition and extreme impulsivity in response to demands may be explained by impairments in executive functioning (Zelazo, Muller, Frye & Marcovitch, 2003). Alternatively, the blanket avoidance response to any demands, even things the child may want to do, may be explained by difficulties in 'seeing the big picture' proposed in the weak central coherence theory of ASC (Frith, 1989), in that any demand is perceived as threatening, regardless of the specific nature of it.

It may also be the case that there are other mechanisms underlying the distinct PDA traits that have yet to be studied. For example, previously it was thought that a lack of accuracy in understanding the emotions of oneself and others was impaired across all individuals with ASC. However, more recent research has argued that this symptom actually occurs heterogeneously across ASC as a result of co-morbid alexithymia (Bird & Cook, 2013; Shah, Hall, Catmur & Bird, 2016), a condition which is not unique to ASC and is present across the wider population (Salminen, Saarijarvi, Aarela, Toikka & Kauhanen, 1999). Thus, although traits or behaviours may be explained by well-formed theoretical arguments, this is not always indicative of a completely comprehensive understanding. If we consider again the concept of equifinality, it may be too reductionist to seek to explain PDA traits which have so far

only been studied in relatively small samples, as occurring in terms of singular underlying pathways or mechanisms.

An alternative explanation for these findings is that children with ASC and high levels of PDA traits may take a more deliberative and imitative route to behaviours when compared to their typically developing and low PDA trait ASC counterparts, evidenced by increased engagement in social mimicry and imitation in this population (Green et al., 2018). Social imitation helps to form social bonds and learn about the world, and is fundamental to forming group memberships (Carpenter, 2006). Imitation can be understood in terms of a catalogue of different behavioural competencies, many of which are reliant on different underlying mechanisms (Jones, 2007). Although previous literature has suggested social imitation and mimicry are impaired across ASC (e.g. Marsh, Pearson, Roper & Hamilton, 2013), this may be a somewhat simplistic assumption which does not fully encapsulate the significant variability between individual presentations.

Thus, behaviours symptomatic of high levels of PDA traits, such as social manipulation or engagement in pretence and fantasy, may result from a more imitative, behavioural learning mechanism, as opposed to occurring more spontaneously as is observed in typically developing children. There may be a lack of awareness on the part of children that they are engaging in these behaviours as they are occurring more simply as a result of mimicry and as a learned response to facilitate the avoidance of demands. This conceptualisation supports the argument that PDA is best explained as a set of reactive symptoms within the context of ASC, or as a response to extreme sensitivity to the social environment (Green et al., 2018). Therefore, although theoretically PDA traits may appear to relate to ToM, there may be many other plausible explanations to explain behaviours.

In contrast, these findings may also be explained with reference to the original sample of children with ASC and PDA traits studied by Newson et al. (2003), where there was an over-representation of girls compared to the non-PDA ASC controls, which may have influenced the description of the presentation. For example, the Pretence and Fantasy subscale in our analysis was the only scale which did not significantly correlate with measures of other PDA traits. Given that engagement in pretence and role-play has been demonstrated to occur more frequently in girls than boys, both in ASC and typically developing populations (Jones & Glenn, 1991; Knickmeyer, Wheelwright & Baron-Cohen, 2008), this trait may have been over emphasised in original accounts of PDA due to the gender distribution between the samples. Thus, although previous research has found that ToM is related to the lack of engagement in pretence in ASC (Chan, Chen, Feng Lee & Chen, 2016), the potential relationship between PDA traits and ToM may have been over-estimated in the original conceptualisation of PDA as the prevalence of symptoms was subject to gender bias.

2.5.3 Clinical Description of PDA

Although PDA traits did not significantly relate to ToM in this sample, there is speculative evidence to support the clinical utility of labelling PDA traits as an important set of behaviours within the autism spectrum. Children in the ASC sample with high rates of PDA traits were more likely to require specialist educational provision than those with low PDA traits. This supports previous findings about the increased level of need in children with high PDA traits (Gore-Langton & Frederickson, 2014; O'Nions et al., 2014). In addition, higher overall levels of emotional and behavioural difficulties, higher levels of conduct problems, internalising behaviours, and externalising behaviours were reported for children with

high PDA traits compared to those with low PDA traits. Further to this, higher levels of communication impairments were reported for children with high PDA traits. Thus, these findings reflect a group of children on the autism spectrum with an increased level of severity in emotional and behavioural difficulties, and communication impairments. This is likely to result in an increased impact on children and families, and an elevated level of need.

In addition, the PDA subscales tested provided necessary insight into how PDA symptoms might hang together as a construct. Although the behavioural profile of PDA has been developed based on clinical accounts (e.g. Gillberg, Gillberg, Thompson, Biskupsto & Billstedt, 2014; Reilly et al., 2014), there has been no evidence from population representative ASC samples of the extent to which traits covary, and whether PDA represents a unitary construct. As mentioned previously, all measures of PDA traits were correlated, with the exception of the subscale measuring engagement in pretence and fantasy; an important facet in descriptive accounts of PDA (Newson et al., 2003). There was no significant relationship between reported scores on this subscale and PDA behaviours reported on the HSQ-ASD, suggesting that the different traits associated with PDA do not necessarily co-occur to the same extent across ASC, or covary with each other. This means that in order to form a comprehensive understanding of PDA traits, and of the behaviours and implications arising from them, the presence of these traits needs to be fully assessed at an individual level as part of the ASC diagnostic process.

2.5.4 Methodological Limitations

Whilst there are a number of plausible explanations for the findings from this study, it is important to considerer these in line with the potential limitations in methodology.

When carrying out multiple correlation analyses, it is necessary to control for the increased risk of type I errors. This is important to consider when interpreting the nominal significance in associations between ToM and some PDA traits, which lacked significance following correction. In this analysis, the Bonferroni correction was applied to the alpha value at which significance was inferred. Although the Bonferroni method is arguably the most appropriate correction for multiple correlations (Curtin & Schulz, 1998), there is contrasting evidence which suggests it can be an over-conservative approach when measures are inter-correlated (Conneely & Boehnke, 2007). It then follows that although the risk of type I error was reduced, the chance of type II error may have been inflated. Therefore, it is difficult to conclude with certainty that there is a complete lack of relationship between ToM and PDA traits, when applying a more liberal correction (e.g. the use of permutation tests) may have resulted in increased significance. In addition, several of the correlations between ToM and PDA traits were approaching significance. If these findings were to be replicated in a larger sample, there may be sufficient power to detect small effects resulting in increased significance.

The use of parent-report methodology also raised a number of potential issues. Although all individual measures reported good validity and reliability statistics, using this method of reporting runs a higher risk of incurring common methods bias (Kamakura, 2010). This bias is especially relevant when studying clinical populations and can only be addressed by using a multi-method, multi-trait approach to data collection. It is likely that parents of children in a clinical study group who are aware that their children are being compared to controls, may be susceptible to bias in reporting, and may over-emphasise behavioural problems compared to parents who are aware their children are being tested as a control group. This effect is likely to be

particularly salient when considering parent-reported difficulties on measures of PDA traits, as many parents report feeling that the severity and impact of behavioural problems associated with PDA are not understood by professionals or the general population (Gore-Langton & Frederickson, 2016).

Further to the more general problems with parent-report measurement tools, ToM is a broad and multi-faceted construct and as such, valid and reliable measurement can be difficult to achieve. Although the ToMI-2 is a validated and sensitive measure (Hutchins et al, 2012), the data collected using the ToMI-2 presented a number of potential issues. Firstly, standard scores were used for analysis and as such, should have reported normal distributions. However, the distribution of two subscales violated assumptions of normality and required transformation. Whilst appropriate for this methodology, transformations can be problematic and result in reduced sensitivity to detect effects (Lo & Andrews, 2015), potentially reducing the likelihood of significant findings using the transformed subscales. Secondly, the scores on the ToMI-2 for the ASC group reported less variability than was observed in initial validation studies (Hutchins et al., 2012). Whilst the ToMI-2 has the sensitivity to detect variation even in low scores, approximately 80% of the scores in the ASC group were at the first percentile, indicating decreased heterogeneity in ToM scores in the sample.

This lack of variability raised questions over whether it is possible to garner a complete understanding of a child's ToM capacity from the use of a parent-report tool only. Whilst parents are thought to be relatively adept at comparing their own understanding with inferences made from the behaviour based understanding of their children, there may be less accuracy in reporting on more advanced reasoning, which often has less observable components (Hutchins et al., 2012). For example, it may be

easy for parents to infer that if they look up at the sky, their child will do so also (e.g. Early ToM). However, it may be more difficult for parents to infer whether a child is able to understand that people have thoughts about others' thoughts (e.g. more advanced ToM). This suggestion is supported by the finding that there was more of an association between the observable, behavioural features of PDA (e.g. as reported by the HSQ-ASD) and ToM, as opposed to some of the slightly more abstract and internal constructs of PDA measured by the EDA-Q.

The lack of variability in ToM scores also led to a consideration of whether the ASC sample in this study was representative of the wider ASC population. Participants were recruited through online platforms and volunteered to take part, with little incentive other than furthering the understanding of PDA traits in ASC. As such, the parents who offered to take part and who completed all measures were likely to have been motivated to give their time for this reason. It is feasible that, as PDA is a relatively under-researched area, parents of children who display challenging traits or behaviours may have been most likely to participate due to a desire to contribute to a better and more publicised understanding of the label. This may have resulted in an ASC sample who displayed elevated PDA traits and more significant behavioural difficulties when compared to a wider ASC population, thereby bringing the generalisability of the findings into question.

Finally, it is important to remember when drawing conclusions that correlation does not infer causation. Further research is needed to ascertain any cause and effect relationship between ToM and traits and behaviours associated with ASC and PDA.

2.5.5 Clinical Implications

Whilst there are several methodological limitations arising from this study, there are still important clinical implications for the findings.

PDA traits in ASC are a relatively under-researched concept. This study contributes important findings to inform a wider understanding of PDA trait presentations in ASC. In particular, the finding that emotional and behavioural difficulties and ASC specific traits and behaviours differ between children with low and high levels of PDA traits, is of some clinical relevance as it contributes to a much-needed understanding of the behavioural symptoms of those who most resemble accounts of PDA. This also has implications for the neurodevelopmental assessment process more generally, where there is currently no requirement for a comprehensive assessment of behaviours relevant to PDA traits. A clear focus on assessment of PDA traits during the ASC diagnostic process would help provide further clarity on the presentation, support more individualised understandings of behaviour, and inform better treatment planning (Green et al., 2018). It would also provide an early understanding of subthreshold traits of PDA, which may be potential risk factors for the development of challenging behaviours and mental health problems.

In addition, the finding that ToM relates to ASC traits but not those observed in accounts of PDA has further clinical implications. The relationship between core ASC traits and ToM provides support for previous literature and provides speculative evidence that ToM may be predictive of ASC behaviours. The lack of relationship between PDA traits and ToM provides evidence to suggest that these traits should be understood differently to those associated with typical ASC presentations. This is of particular relevance in the assessment and diagnosis of ASC, and for the development of individual and person-centred care planning. For example, the findings provide support to the suggestion that ToM training programs may be helpful in reducing the core social and communication difficulties observed in ASC (e.g. Fisher & Happe, 2005; Fletcher-Watson, McConnell, Manola & McConachie, 2014); but also highlight

that this may not be an effective intervention to manage the social difficulties associated with PDA presentations.

However, it is also important to consider that ToM scores did not differ between the ASC-PDA and ASC+PDA groups. One explanation for this is that there may be a shared vulnerability pathway between ToM difficulties and behavioural learning or emotion regulation mechanisms, which impacts the presence of PDA traits and behaviours. This would imply that whilst ToM does not differ with presentations of PDA traits, behaviours associated with PDA may emerge in relation to a neurodevelopmental problem with connectivity. Thus, there is evidence to support the suggestion postulated by Green et al. (2018); that clinically, PDA traits should be described in terms of underlying mechanisms related to ASC *and* the overlap in symptomology with additional diagnoses, dimensional descriptions of behaviour, and contributing risk and protective factors.

2.5.6 Implications for Further Research

The findings and methodological limitations of this study give rise to several implications for further research. The finding that ToM accounts for a significant amount of the variance in core ASC traits but not for PDA traits, contributes to a better understanding of these behaviours and can be used to inform relevant strategies to support individuals with ASC. However, given the methodological limitations and nominal significance observed between some facets of ToM and some PDA traits, further research is needed to draw definitive conclusions on this relationship. One area of focus should be the use of more comprehensive ToM measurement, comprising observations, tests and multi-informant reports (Wellman et al., 2001) in a larger and more representative sample of children, resulting in improved power to detect effects. This could also facilitate an investigation of other facets of ToM, e.g. cognitive and

affective, and exploration of whether any association with PDA traits emerges in relation to these subcomponent processes.

In addition, there is still a need for further evidence on the validity of PDA as a distinct construct, as opposed to a presentation which could be described by a diagnosis of ASC plus another comorbid disorder with symptoms of challenging behaviour (e.g. ASC+ODD; Green at al., 2018). Further to this, a more developed understanding of the potential underlying mechanisms driving PDA traits and behaviours in needed. To support this, further research should focus on the continued development of comprehensive, multi-informant and clinically relevant measurement tools for PDA traits and the investigation of other plausible mechanisms underlying these behaviours. This could begin by exploring processes implicated in ASC presentations, for example, central coherence and executive functioning (investigated in the other paper of this joint project). The ideas proposed by Green et al. (2018) could also be investigated in more detail: that PDA may occur in relation to an intolerance of uncertainty, altered sensory perception, low generativity and predictability, and emotional dysregulation. Further research in this area is crucial to inform the development of appropriate behavioural management strategies to support children with high PDA trait presentations at home and in educational settings.

2.5.7 Conclusions

The findings from this study contribute to the existing body of literature exploring PDA traits and behaviours in ASC. The distinct behavioural profile of children with ASC and high PDA compared to those with low PDA traits provides support for previous research (e.g. O'Nions et al, 2014) and evidence to support the use of PDA as a clinically relevant and valid label to describe these symptoms as they occur within the autism spectrum.

With regards to the relationship between ToM and traits relevant to ASC and PDA, ToM emerged as a significant predictor of ASC traits, with lower ToM scores associated with increased reports of ASC traits. In contrast, there was not significant evidence to suggest an association between ToM and PDA traits. However, given the methodological limitations outlined, further research is needed to draw more definitive conclusions on this relationship and to provide further hypotheses to explain the underlying mechanisms behind PDA behaviours. This would further inform the understanding of PDA traits in ASC and contribute to development of comprehensive assessment schedules and more individualised behavioural management strategies.

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Part 3: Critical Appraisal

3.1 Introduction

This appraisal builds upon and outlines some of the insights and understandings I have gained throughout the process of conducting this literature review and research study. Considerations on the design, methodology and results of the research study will be discussed. Broader conceptual issues with measurement tools will then be considered, with a focus on the difficulty of measuring theory of mind (ToM) reliably in this research. The appraisal will conclude with a general discussion on the experience of carrying out this research.

3.2 Design and Measures

Whilst planning and conducting this thesis, a number of potential issues with design and methodology came to light, leading to broader considerations of how these impact on research more generally.

3.2.1 Planning a Joint Project

There were a number of deliberations and decisions with regards to planning a joint thesis which needed to be made prior to submitting an application for ethical approval. Most notably, carrying out a joint project gave rise to two different sets of hypotheses, both requiring data collection from the same set of participants. This meant it was necessary to compromise on the measures included for use, and to only use one measure to assess each construct. As we were only able to use single informant-reports on a single measurement tool, our results were more susceptible to common methods bias, resulting in limitations to the reliability and validity of our methodology. However, although working on a joint project meant the information we were able to collect was slightly restricted, it was invaluable for a number of reasons.

Firstly, it allowed us to share the burden of recruitment and respond more quickly to interested participants. This increased the target population we were able to reach and the speed at which we were able to enrol participants into the study. Anecdotally, we noticed that participants were more likely to complete the questionnaires when they were sent with minimum delay, meaning we were able to minimise the attrition rate of participants. Secondly, joint working facilitated the sharing of ideas and provided alternative perspectives to inform the research design. This contributed to a better-planned study as potential limitations were more likely to be identified and addressed. Finally, it was reassuring to have someone to discuss the more stressful aspects of data collection and recruitment with, and to share the motivation of the project with.

3.2.2 Measure Selection for the Empirical Paper

In addition to constraints on measure selection arising from working on a joint project, there were other important elements of measure selection which needed to be considered during the planning stage of the study.

As we were studying a clinical ASC population and control group, we had to exclude the use of certain measures which were not deemed appropriate or had not been validated for use in children with neurodevelopmental conditions. Whilst our measure of emotional and behavioural difficulties (SDQ; Goodman, 2001) was deemed appropriate for use in ASC and typically developing samples (Simonoff et al., 2012), there were questions raised over the comparison of subscale scores between groups. The SDQ is most commonly analysed in terms of its five subscales: Emotional, Conduct, Hyperactivity, Peer Relationships and Prosocial Behaviours. However, there is emerging evidence to suggest that whilst these subscales are appropriate for screening for disorders (Van-Roy, Veenstra & Clench-Aas, 2008), they have limited

sensitivity when applied to a typically developing population. Instead, broader subscales measuring Internalising and Externalising behaviours are recommended for use (Goodman, Lamping & Ploubidis, 2010), making valid comparisons between groups difficult.

In addition, we also had to ensure we selected measures of ASC traits and behaviours which were appropriate for use in typically developing populations. As a result of this, measures differed in sensitivity and specificity between groups, resulting in an increased potential for type I or II errors in the findings. Thus there were methodological difficulties in using not only the same measure, but the same subscales to measure traits in both clinical and control populations.

We also had to consider how best to collect a vast amount of information from participants, without increasing the attrition rate of our study. As mentioned previously, this was a difficult balance to achieve. Ideally, we would have used additional measures to give a broader overview of behavioural difficulties and how these differ between an ASC and non-ASC sample, and between children with low and high PDA trait presentations in ASC. We would also have considered the feasibility of including a task-based assessment of ToM for children in our ASC sample, an important and well-validated component of ToM measurement (Wellman, Cross & Watson, 2001). However, we were mindful that our participants were already giving up a significant amount of time to complete questionnaires, with no immediate or tangible incentive. As such, we chose to limit the measures used, possibly at the expense of discovering more in-depth information about a relatively under-researched clinical presentation.

3.2.3 Problems with Informant-Report Measurement Tools

Another issue to consider when designing the empirical study was the potential for limitations arising from the sole use of informant-report measurement tools.

When conducting the literature review, it became apparent that there were numerous informant-report tools available to measure one construct (aggression), all of which reported good validity and reliability statistics. However, with such variability in composition and specific items included, there were reservations about the subjective nature of these measurement tools. In addition, although there are many validated informant-report tools which are appropriate to use to measure a complex construct like aggression, it is difficult to infer with any clarity whether it is possible to draw conclusions from comparisons between measurement tools. Whilst scores on most measures correlate highly with one another (e.g. Pellegrini & Bartini, 2000), it is arguable that different measures may tap different facets of the same construct (e.g. focus more on measuring relational aggression, as opposed to physical aggression). This implies that whilst measurement tools may refer to an overall measure of one construct, they may in fact be measuring different traits and behaviours occurring under the same conceptual umbrella (Archer & Coyne, 2005; Dodge & Coie, 1987).

There is also a possibility that different measures may bias different styles of response, based on the personal style or preference of the responder. This is supported by evidence which suggests descriptions or interpretations of child's behaviour may be informed by the individual traits or characteristics of the responder (e.g. Najman et al., 2001). Research has demonstrated that abusive parents rate their children as having significantly more conduct problems than non-abusive parents, and in comparison to independent observations of their child's behaviour (Reid, Kavanagh & Baldwin, 1987). This implies that parents can be biased in identifying and drawing inferences from their child's behaviour, based on their own behaviours and the relationship they

have with their child. For example, parents who score higher on tests of hostile attribution bias may be more likely to rate their children as more aggressive, or as having higher rates of socially manipulative behaviours. Whilst the use of multiple methods of measurement were beyond the scope of this thesis, there must be some consideration of the limitations of single-informant-report measurement when interpreting our findings.

3.2.4 ToM Measurement

Ultimately, the limitations of informant-report measurement tools were likely to have been most salient for the measure of ToM used in the empirical study.

ToM is most often assessed directly with children, either on false belief or unexpected contents tasks, or through other language based assessments (Wellman et al., 2001). It is widely agreed that the more simplistic false-belief measurement methods are problematic due to their pass or fail nature – they imply that ToM is something you either have or you don't (Hutchins, Bonazinga, Prelock & Taylor, 2008). A recent systematic review of ToM measures suggests there are a number of valid and reliable measures which go beyond simple false-belief assessment (Ziatabar Ahmadi, Jalaie & Ashayeri, 2015). The review recommends the selection of a measure based on the reported psychometric properties for the population in question. That the ToMI-2 reports good levels of validity and reliability for both typically developing and ASC populations (Greenslade & Coggins, 2016; Hutchins, Prelock & Bonazinga, 2012) should therefore make it a good candidate for use. However, there are limitations to the extent that any informant-report measure is able to fully assess ToM. There is general agreement that comprehensive assessment needs to be comprised of a battery of tasks tapping different facets of the socio-cognition at different levels of complexity (Hughes et al., 2000; Tager-Flusberg, 2001).

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Again, as discussed previously, there may also be confounding factors with relation to individual informant characteristics. The informants in our study were all parents, and mostly mothers (98%). There is evidence to suggest that a mother's conversational preferences and use of mental state language in infancy directly impact their child's ToM development (Meins et al., 2002; Peterson & Slaughter, 2003; Ruffman, Slade & Crewe, 2002). Therefore, it is reasonable to suggest that a mother's mind-mindedness and ability to use mental state language may impact the extent to which they are able to identify these skills in their own child. This implies that the ToM ratings for the children in our study could have been subject to biases. As we did not collect data on parental mind-mindedness or mental state term ability, we were not able to assess or control for this, potentially impacting the validity and reliability of the results.

Thus, given the well-known and widely researched improvements to validity and reliability when using multi-informant, multi-method approaches to complex assessment (Campbell & Fiske, 1959; Merrell, 2008; Tahiroglu et al., 2014), there is an argument to suggest that a single-informant measure of ToM may never be fully adequate to comprehensively assess the concept. Had we been able to include a direct assessment of ToM alongside informant-report measurement, this may have allowed for a more in-depth understanding of the ToM profile in ASC, both for children with low and high PDA trait presentations.

3.3 Process of the Empirical Study

In addition to considerations of the design of the empirical paper, and problems with informant-report measurement tools, I also reflected more broadly on the

challenges and learning points which arose during the process of conducting a research study.

3.3.1 Recruitment

One of the biggest challenges faced by any research study is likely to be problems with recruitment (Patel, Doku & Tennakoon, 2003). This was certainly the case when we started recruiting to this study. Between November 2017 and January 2018 we were only able to recruit 27 participants, less than a third of our desired sample size. At this point, we increased recruitment efforts and asked our supervisors to help publicise the study via social media platforms. This made a difference and we were inundated with requests to participate, quickly reaching our desired sample size of 90 participants.

Whilst helpful in achieving our designated sample size, using such targeted recruitment methods, snowballing techniques, and the reliance on social media, gave way to a host of other problems. We found that the majority of people contacting us about the study were parents of children with a diagnosis (or suspected diagnosis) of PDA. As we were trying to capture data from children with a broad spectrum of PDA traits in ASC, we needed to ensure our ASC sample was not just comprised of children with high levels of PDA traits. This meant we had to monitor PDA traits during recruitment and exclude potentially eligible participants in the study. Whilst this likely resulted in a more representative ASC sample, controlling recruitment in this way was arguably not the most methodologically sound approach.

On reflection, we wondered more about the way in which parents of children who had been given a diagnosis (or suspected diagnosis) of PDA from professionals responded to our research. We noted the high level of interest in PDA communities online, and that there seemed to be increased rates of peer support available. We

wondered whether this was, in part, due to the relative lack of research and clinical understanding of PDA traits, and that the term is not included in diagnostic and statistical manuals. In addition, parents of children identifying with descriptions of PDA appeared much more interested in, and motivated to complete our research than those reporting more typical presentations of ASC. We thought further about the demands placed on parents in managing the more challenging aspects of PDA traits in ASC, especially for those who do not feel that a diagnosis of ASC fully captures their child's difficulties. We considered that perhaps these parents were the most motivated to contribute to a wider understanding of difficulties associated with the PDA presentation, in the hope it would raise awareness and acceptance of the label. We also reflected that, whilst seen as reductionist by many, appropriate diagnosis and labelling of symptoms can provide a much needed understanding of a presentation for some, and reduce the stigma associated with more difficult behavioural symptoms.

3.3.2 ASC Sample

Another interesting observation made during the process of this research study was that there were seemingly higher rates of homogeneity in ASC and PDA related behaviours within our high PDA trait sample, compared to those with low PDA traits. It is widely agreed that there is significant heterogeneity in ASC, and the condition is thought by some to be characterised by this variability (e.g. Newschaffer, Fallin & Lee, 2002; Towgood, Meuwese, Gilbert, Turner & Burgess, 2009), so the finding that this appears reduced in autistic children with high PDA traits is of note. However, it is important to consider this finding in line with methodological limitations in drawing conclusions from such a small sample, and with reference to the aforementioned potential impact of using parent-report to measure challenging behaviours which do not have a widely accepted clinical label. With this in mind, we considered that the

observed homogeneity in our high PDA traits group may have occurred as a result of reporter bias, in that parents of children with high PDA traits may have been those most striving for an explanation of the behaviour, and so be more susceptible to over-report behavioural difficulties.

In relation to the observed homogeneity in our ASC+PDA sample, we considered more broadly whether the ASC sample as a whole were truly representative of a typical ASC population, or whether they reported increased rates of PDA traits and behavioural difficulties. When comparing our ASC-PDA group to O'Nions et al's. (2014) sample of children with ASC who did not display disruptive behaviours, we found increased levels of conduct problems in our sample. There were also higher mean scores on the EDA-Q for our ASC-PDA *and* ASC+PDA groups when compared to the ASC samples studied by O'Nions et al. This suggests that the ASC sample in this study may have had over-elevated rates of PDA traits than are observed in the wider ASC population. This has implications for the generalisability of the results.

3.4 Results of the Empirical Paper

Overall, the empirical study found evidence to suggest ToM is related to core ASC traits, but not to PDA traits, and to support the idea that behaviour differs in relation to high and low levels of PDA traits in ASC. In addition to the observations and insights drawn from the process of conducting the literature review and empirical study, the results of the empirical paper led to a number of further considerations.

3.4.1 Data and Distributions

As previously mentioned, the measures used in this study needed to be appropriate and valid for use in a clinical ASC and a non-ASC control sample. Perhaps as a result of this, the scores on the ToMI-2 for the sample as a whole violated the

assumptions of normality, reporting bimodal distributions and significant levels of skew and kurtosis. Distributions for the whole sample, and the ASC sample alone are shown in Appendix N. It was therefore not possible to apply an appropriate transformation to the data, meaning non-parametric analyses were used for standard scores on measures which should have reported normal distributions. This limited the extent to which comparisons could be inferred between parametric and non-parametric tests, and the conclusions we were able to draw from results.

3.4.2 Publication Bias

The experience of conducting this study also led me to reflect on the effects of the researcher's response to findings, and the potential disappointment of reporting null findings or those which do not provide support for study hypotheses. The findings from the empirical paper, that ToM did not significantly relate to measures of PDA traits in ASC, were undoubtedly interesting and have many clinical implications. However, my first reaction to this result was an initial disappointment that I had not been able to prove my hypotheses. This led me to consider the wider issue of publication bias in academic research.

Publication bias occurs when the likelihood of findings from a study being disseminated is affected by the outcome itself, with the idea that null findings are less likely to be reported (Dickersin et al., 1987; Kicinski, Springate & Kontopantelis, 2015). This becomes particularly problematic when literature reviews or syntheses are conducted as it raises the probability of incurring a type I error (Rothstein, Sutton & Borenstein, 2005). The risk of publication bias is likely to be reduced by conducting better-powered studies, enhancing research procedures and considering interpretations of results with more scrutiny (Ioannidis, 2005). However, if an initial response to the outcome of a study is one of disappointment, it therefore follows that there is likely to

be less motivation to go through the effort of publishing the findings (Easterbrook et al., 1991). This is important to consider in tackling publication bias as it belies an issue beyond that of simply improving research designs, and suggests null findings need to be reframed as a crucial and important contribution to the literature on any given subject.

3.5 Conceptual Issues

Whilst conducting the literature review and empirical paper, I reflected more broadly on the difficulties in conceptualising broad constructs such as ToM in research. I also considered difficulties arising from researching PDA more specifically and my own experience of this.

3.5.1 Conceptualising ToM

When measuring broad and multi-faceted concepts such as ToM, there are important considerations to be made with regards to how it is conceptualised as a construct. As highlighted in the literature review and empirical paper, there is an increasing body of research supporting the need for a distinction between cognitive and affective ToM (Kalbe et al., 2010; Shamay-Tsoory & Aharon-Peretz, 2007). Studies have proffered evidence to suggest these concepts can occur independently of one another, and develop at different rates during different developmental timeframes (Sebastian et al., 2012). One of the notable limitations identified in the literature review was that so few studies included a distinction between cognitive and affective ToM. It therefore follows that the inability of the empirical paper to conceptualise ToM in terms of measuring both cognitive and affective facets of ToM, limits the depth and brevity of understanding we are able to draw from the findings.

As of yet, there is not an informant-response tool which measures these distinct aspects of ToM and as such, it was not within the scope of this thesis to do so. However, given the widely reported heterogeneity in ASC, and the limited number of empirical studies examining the distinction in cognitive and affective ToM in an ASC population (Kim et al., 2016), this level of depth may have provided valuable and unique insights into the relationship between ToM and key behaviours in ASC. This is especially salient when we consider the suggestion that perhaps it is the understanding of belief and cognitive states that poses a challenge in the ASC population, as opposed to more affective difficulties understanding desire and emotion (Peterson, Wellman & Liu, 2005).

It is also important to consider the role of mediating and moderating factors on the relationship between ToM and behaviours. It can be difficult to conceptualise ToM fully without due attention to these factors, and is therefore problematic that we did not collect data which would allow us to control for known correlates of ToM. This is particularly important for the role of language ability as it is widely acknowledged that children's ToM understanding relates to this (Milligan, Astington & Dack, 2007). Given that one of the symptomatic PDA traits described by Newson, Marechal & David (2003) was language delay associated with increased passivity, and an improved ability to "catch-up" than is observed in more typical ASC presentations (Green et al., 2018), it would have been particularly interesting to explore the role of language further. Whilst we controlled for more general cognitive impairments by excluding children with moderate to severe learning disabilities, we did not include any way of reporting the presence of a specific language impairment or disorder. Although it is difficult to find a parent-report measure of language ability validated for use, we

should have considered how to control for this when designing the study. This was an important omission and raises questions for further research in this area.

3.5.2 Researching PDA

Another area of contention highlighted during the design and process of the empirical paper was the difficulty in carrying out research into presentations which have not been classified under nosological criteria. When this research project began, our brief was to focus on investigating the underlying cognitive processes and mechanisms associated with behaviour in children meeting criteria for PDA. At this time, we were thinking of PDA as a distinct and categorical diagnosis; a separate syndrome under the conceptual umbrella of ASC. From this theoretical standpoint, we planned to conduct between-group analyses to investigate differences between children with ASC with and without PDA, and non-ASC controls. However, during the process of conducting the thesis, new findings were published suggesting there is not sufficient evidence to argue for PDA as a standalone diagnosis (Green et al., 2018), suggesting that PDA traits occur dimensionally across presentations of ASC. This led us to question whether solely conducting between-group analyses was appropriate.

With this in mind, PDA traits were explored continuously across the ASC sample as a whole. However, making changes to the design and data analysis plan at a relatively late stage in the research process posed a challenge. To begin with, I struggled to change the way I had initially conceptualised PDA. This led me to think in more detail about the difficulty conducting research in an area where there is limited literature to inform an understanding of a presentation. I also considered that when a concept is relatively under-researched and in its early inception, there are likely to be areas of contention before a joint understanding can be agreed upon and researched further (although arguably, it can be difficult to reach a shared understanding for even

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some of the most well-researched conditions, for example, the controversy surrounding diagnoses of Attention Deficit Hyperactivity Disorder). With this in mind, I considered that our initial plan to focus solely on between-group analyses was perhaps premature and that it would have been more appropriate to frame the research as more loosely exploratory from the outset (Shields & Tajalli, 2006).

I also reflected on the difficulty arising from studying a relatively new area that I did not, initially, feel I had relevant clinical experience of. Although there is relatively comprehensive information available to describe the behavioural symptoms of PDA from both professionals and parents, this felt rudimentary compared to the depth of understanding which can be gained from working directly with a population. However, as my understanding of PDA moved from a more categorical framework to occurring dimensionally in ASC, I thought more about my own experiences working with children whilst on placement in a neurodevelopmental team. This helped me to form a clearer understanding of PDA traits in my own mind, as I was able to consider some of the more atypical behavioural traits observed in children, and how these might fit with existing descriptions of PDA. I also reflected further on how these traits differed so significantly between individuals. Having a better informed understanding of how PDA traits might present in children helped to increase my confidence in researching in this area and informed a more comprehensive understanding of the presentation.

3.6 Conclusions

Throughout the process of conducting this research, I have become increasingly aware of the complexity of ASC. This has been particularly relevant in terms of how it is conceptualised, and the idea that both ToM and PDA traits in ASC need to be conceptualised as occurring dimensionally across ASC, as opposed to a

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categorical presentation. This, alongside the well-documented heterogeneity in presentation, make ASC a difficult area to study. This is perhaps reflected by the extreme disparity between the economic impact of the condition and low levels of research spending (Buescher, Cidav, Knapp, & Mandell, 2014). The expertise of my supervisors has therefore been instrumental in facilitating my understanding in this area.

I have also considered the multiple areas of issue arising from the measurement of cognitive and behavioural constructs, often by way of informant-report measurement tools. I have discussed difficulties with the use of numerous measurement tools to measure one behavioural construct, and the validity of comparisons between these measures. I have considered the reliability and validity of informant-report measurement tools more generally, and specifically when measuring socio-cognitive concepts which are not directly observable. Finally, I have considered the importance of measuring complex constructs such as ToM using a multi-method, multi-informant measure, and the difficulty of achieving this in relatively small scale research studies.

I have learnt a great deal about PDA traits in ASC. However, there is still an enormous amount of research necessary to improve a wider understanding of this constantly evolving area. Future studies should further investigate behavioural presentations associated with PDA traits and how these are measured. There should also be a focus on exploring the underlying cognitive processes and mechanisms implicated in PDA traits in ASC, using a variety of measurement tools and methods. This would contribute to a better understanding of PDA traits in ASC and help to develop vital knowledge to inform the clinical assessment and utility of the term.

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Appendix A

Individual contributions to a joint project

This project was conducted in collaboration with another Trainee Clinical Psychologist from University College London (UCL), Anna Goodson. Anna and I were both responsible for creating basic ideas and research questions for the project. We each took sole ownership of more specific hypotheses and, with the support of our supervisors (Dr Will Mandy and Dr Liz O'Nions), created more in-depth arguments to support these.

Recruitment and data collection responsibilities were shared between Anna and me. We both independently contacted separate organisations via email and social media to request our study was advertised, and were both responsible for enrolling participants into the study and providing a point of contact for any queries.

The data for each of our papers was collated and analysed independently. As such, we were both responsible for the write-up of two completely separate empirical papers, both exploring different hypotheses regarding the underlying mechanisms of PDA traits in ASC.

Appendix B

Research Ethical Committee Approval Letter

UCL RESEARCH ETHICS COMMITTEE
ACADEMIC SERVICES

🏛 UCL

24th February 2017

Dr William Mandy
UCL Division of Psychology and Language Sciences

Dear Dr Mandy

Notification of Ethical Approval
Re: Ethics Application 10193/001: Exploring demand avoidance in children with and without Autism

I am pleased to confirm in my capacity as Chair of the UCL Research Ethics Committee (REC) that your study has been ethically approved by the REC until **30th September 2018.**

Approval is subject to the following conditions:

Notification of Amendments to the Research
You must seek Chair's approval for proposed amendments (to include extensions to the duration of the project) to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature. Each research project is reviewed separately and if there are significant changes to the research protocol you should seek confirmation of continued ethical approval by completing the 'Amendment Approval Request Form':
http://ethics.grad.ucl.ac.uk/responsibilities.php

Adverse Event Reporting – Serious and Non-Serious
It is your responsibility to report to the Committee any unanticipated problems or adverse events involving risks to participants or others. The Ethics Committee should be notified of all serious adverse events via the Ethics Committee Administrator (ethics@ucl.ac.uk) immediately the incident occurs. Where the adverse incident is unexpected and serious, the Chair or Vice-Chair will decide whether the study should be terminated pending the opinion of an independent expert. For non-serious adverse events the Chair or Vice-Chair of the Ethics Committee should again be notified via the Ethics Committee Administrator within ten days of the incident occurring and provide a full written report that should include any amendments to the participant information sheet and study protocol. The Chair or Vice-Chair will confirm that the incident is non-serious and report to the Committee at the next meeting. The final view of the Committee will be communicated to you.

Final Report
At the end of the data collection element of your research we ask that you submit a very brief report (1-2 paragraphs will suffice) which includes in particular issues relating to the ethical implications of the research i.e. issues obtaining consent, participants withdrawing from the research, confidentiality, protection of participants from physical and mental harm etc.

Yours sincerely

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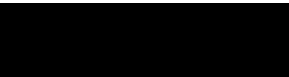
Professor Michael Heinrich
Interim Chair, UCL Research Ethics Committee

Cc: Anna Goodson & Ellie Bishop

Academic Services, 1-19 Torrington Place (9th Floor),
University College London
Tel: +44 (0)20 3108 8216
Email: ethics@ucl.ac.uk
http://ethics.grad.ucl.ac.uk/

Participant Information Sheet – displayed on Qualtrics

Exploring Demand Avoidance in Children with and without Autism

Thank you for expressing an interest in taking part in our study (UCL Ethical Approval Ref: 10193/001)

You should have already received a copy of this information sheet but please check that you have read it carefully before continuing.

Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and feel free to discuss with others if you wish. If there is anything that is not clear, or if you would like further information, please send us an email (e.bishop@ucl.ac.uk / anna.goodson.15@ucl.ac.uk).

What is the research about?
This research is exploring demand avoidance behaviour in children. Having a child who struggles with demand avoidance behaviours can be stressful and challenging at times. Children with demand avoidance difficulties have some of the highest rates of exclusion from schools and education. By taking part in this study, you are helping contribute to an increased understanding of the cognitive processes in demand avoidance. This could help raise awareness, inform behavioural management strategies for children, develop support for parents, and increase the availability and understanding of these.

What does taking part involve?
There are two stages to participation in this study. Firstly, all participants will be asked to complete a short set of questionnaires. You will be taken to this part of the study once you have finished reading the information on this page and given your consent to participate.
Secondly, some participants will be asked to complete some follow-up questionnaires about their child's behaviour. These can be done online or on paper and should take approximately one hour. You do not need to complete all the questionnaires at one time and will be able to save your progress so you can return to it at a more convenient time.
After completing the questionnaires, to thank you for your participation you will be given the chance to enter a prize draw to win an Amazon voucher, ranging in value from £10 to £50.

What will happen to my information?
All information collected about you during the course of the study will be kept strictly confidential and stored in secure University College London (UCL) premises. Your name and contact details will be stored separately from the data collected. All

information will be kept securely according to the requirements of the Data Protection Act 1998.

It is likely that the results of this study will be published, but any published outcomes will remain strictly anonymous. Only group results will be presented and no individual will be discussed. Identifiable information (such as name or date of birth) will not appear on any publications or reports about this research. If any work is to be published, you will be notified of this and able to request a copy.

Do I have you take part?

No. It is up to you to decide whether or not to take part in this study. In other words, this is voluntary. If you do decide to take part you are still free to stop your participation at any time and have any research data withdrawn without giving a reason.

Having read this information, do you wish to continue with this study?

Appendix D

Participant Consent Form – displayed on Qualtrics

By continuing with this study, I agree that:

- I have read the notes written above and the Information Page, and understand what the study involves.

- I understand that if I decide at any time that I no longer wish to take part in this project, I can notify the researchers and withdraw immediately.

- I consent to the processing of my personal information for the purposes of this research study.

- I agree that my data, after it has been fully anonymised, can be shared with other researchers.

- I understand that such information will be treated as strictly confidential and handled in accordance with the provisions of the Data Protection Act 1998.

- I consent to the above and agree to participate in this study.

By continuing with this study, you consent to all of the above.

○ I consent to participate in this study

○ I do not consent to participate in this study

Research Advertisement

Participant email template – phase one

Dear X,

Thank you for your interest in our study. It would be great if you could participate in our research. All the information you need should be below, but if you have any questions or comments regarding the questionnaires, please just email myself or Anna (copied in).

This study involves completing an online questionnaire about your child who is aged between 6-11 years old. All the answers you provide must be related to just one of your children. Based on your responses to these questions, you might be asked to complete some further questions. We will get back in touch with you to let you know if this is the case.

Below is a participant code. This is unique to you and needs to be entered into the online questionnaire. There is also a link which will take you directly to the online questionnaire. It is important that you try to complete the questionnaire in one sitting to ensure it saves all your responses. Therefore, it is best to start the questionnaire when you have about 20 minutes of free time.

If you do not wish to complete the questionnaire online, and would prefer a paper version please let us know by replying to this email with your postal address and we will send them to you with a stamped, addressed envelope for you to return them.

Participant Code:

Password:

Click below for the study link (you may need to hold ctrl + click the link)
Phase 1: Exploring Demand Avoidance in Children with and without Autism

We'll send you reminders every week to complete the questionnaires, but if you want to opt out at any point please just let us know and we'll stop! Additionally, if you know anyone else who might be interested in participating, please feel free to pass our details on.

Many thanks again for your interest and support in our research,

Participant email template – phase two

Dear X,

Thank you very much for completing the first phase of our research study! We appreciate the time and thought taken.

Based on your responses to the first set of questionnaires, we would be very grateful if you could complete the second phase of our study. This is done in **two parts** and it's very important you complete both.

Below is a participant code. This is unique to you and needs to be entered when prompted. There are also **two links** which will take you directly to the online questionnaires. It is important that you try to complete the questionnaires in one sitting to ensure it saves all your responses. Therefore, it is best to start the questionnaires when you have about 15 minutes of free time.

Participant Code:

Password:

PART 1 (you may need to copy and paste the link into your browser window)
https://www.theoryofmindinventory.com/professionals/caregiver-assessment/

Click below for PART 2 link
Phase 2: Exploring Demand Avoidance in Children with and without Autism

Please follow both links separately, as the questionnaires are different!

Again, thank you very much for your help and interest in our study. If you have any questions please do not hesitate to contact either Anna Goodson (anna.goodson.15@ucl.ac.uk) or Ellie Bishop (e.bishop@ucl.ac.uk).

Demographic Questionnaire – displayed on Qualtrics

Please answer the following questions about YOURSELF

1. *Date of Birth (year)*

2. *Gender*

○ Male

○ Female

○ Prefer not to say

3. *First Language*

4. *Ethnicity*

○ White British / White Other

○ Asian British / Asian Indian / Pakistani / Chinese / Other

○ Black British / Black African / Black Caribbean / Other

○ Mixed (please specify [   ]

○ Other (please specify) [   ]

◉ Prefer not to say

5. *Does your child live with you?*

○ Yes - all of the time

○ Yes - some of the time

○ No

Please answer the following questions about YOUR CHILD

1. *Date of Birth (month/year)*

Month [▼]

Year [▼]

2. Gender

○ Male

○ Female

○ Prefer not to say

3. First Language

4. Ethnicity

○ White British / White Other

○ Asian British / Asian Indian / Pakistani / Chinese / Other

○ Black British / Black African / Black Caribbean / Other

○ Mixed (please specify) [____]

○ Other (please specify) [____]

○ Prefer not to say

5. *Please indicate whether your child has ever received one of the following Autistic Spectrum Disorder diagnoses:*

○ Autism Spectrum Disorder (ASD) / Autism Spectrum Condition (ASC)

○ Autism/Autistic Disorder

○ Asperger's Syndrom/Asperger's Disorder

○ High-Functioning Autism

○ Pervasive Developmental Disorder - Not Otherwise Specified (PDD-NOS)

○ Other (please specify) [____]

○ None of the above

6. *If yes to the previous question, where/from whom was the diagnosis received?*

○ Child and Adolescent Mental Health Service (CAMHS)

○ Child and Adolescent Psychiatrist

○ Clinical Psychologist

○ Educational Psychologist

○ Paediatrician

○ Speech and Language Therapist

○ Other (please specify) [＿＿＿＿＿]

○ × I don't know

○ × N/A

7. *Has your child ever been given any of the following:*

○ A diagnosis of PDA (Pathological Demand Avoidance)

○ A diagnosis of demand avoidant traits/PDA features

○ Suspected PDA but not clinically diagnosed

○ None of the above

○ I don't know

8. *If yes to the previous question, who gave your child the diagnosis?*

☐ Child and Adolescent Mental Health Service (CAMHS)

☐ Child and Adolescent Psychiatrist

☐ Clinical Psychologist

☐ Educational Psychologist

☐ Paediatrician

☐ Speech and Language Therapist

☐ Other (please specify) [＿＿＿＿＿]

☐ × I don't know

☐ × N/A

9. What type of school does your child attend?

○ Mainstream school

○ Special school for children with Autism

○ Special school for children with Learning Disabilities

○ Specialised unit within a mainstream school

○ Home school

○ My child is not in education

○ Other (please specify) [   ]

○ × I don't know

10. Has your child been diagnosed with any of the following specific learning disabilities/difficulties? Please select one or more of the options below:

☐ Mild Learning Disability

☐ Moderate Learning Disability

☐ Severe Learning Disability

☐ Profound and Multiple Learning Disability (PMLD)

☐ Dyslexia

☐ Dyscalculia

☐ Dyspraxia

☐ Dysgraphia

☐ Attention Deficit Disorder (ADD)

☐ Attention Deficit Hyperactivity Disorder (ADHD)

☐ Other (please specify) [   ]

☐ None of the above

☐ × I don't know

Appendix H

Childhood Autism Spectrum Test (CAST; Scott et al, 2002)

Removed to comply with copyright

<u>Appendix I</u>

Strengths and Difficulties Questionnaire (SDQ; Goodman, 2001)

Removed to comply with copyright

Appendix J

Extreme Demand Avoidance Questionnaire (EDA-Q; O'Nions et al, 2014)

Removed to comply with copyright

<u>Appendix K</u>

Home Situations Questionnaire – ASD (HSQ-ASD; Chowdhury et al, 2015)

Removed to comply with copyright

Appendix L

Theory of Mind Inventory-2 (ToMI-2; Lerner, Hutchins & Prelock, 2011)

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Appendix M

EDA-Q Subscale Development

Social Manipulation

1. Item 5: Tells other children how they should behave, but does not feel these rules apply to him/herself.
2. Item 11: Good at getting round others and making them do as s/he wants.
3. Item 16: Knows what to do or say to upset specific people.
4. Item 17: Blames or targets a particular person.
5. Item 21: Uses outrageous or shocking behaviour to get out of doing something.
6. Item 23: Social interaction has to be on his or her own terms.

Total Score = (Items 5 + 11 + 16 + 17 + 21 + 23)/6

Reliability Analysis:

	Cronbach's alpha
Controls	α=.801
ASC	α=.816
Overall	α=.902

Overall Inter-Item Correlation Matrix

	Item 5	Item 11	Item 16	Item 17	Item 21	Item 23
Item 5		.620	.512	.591	.724	.755
Item 11			.524	.512	.670	.650
Item 16				.679	.577	.478
Item 17					.607	.638
Item 21						.561
Item 23						

Overall Item Total Statistics

	Scale M if item deleted	Scale variance if item deleted	Corrected item-total correlation	Squared multiple correlation	Cronbach's Alpha if item deleted
Item 5	6.79	24.170	.787	.703	.877
Item 11	6.62	26.534	.718	.573	.887
Item 16	6.80	27.663	.656	.520	.896
Item 17	6.94	25.781	.728	.611	.885
Item 21	7.20	25.731	.762	.655	.881
Item 23	6.38	24.557	.753	.681	.882

Pretence and Fantasy

1. Item 6: Mimics adult mannerisms and styles (e.g. uses phrases adopted from teacher/parent to tell other children off).
2. Item 8: Takes on roles or characters (from TV/real life) and 'acts them out'.
3. Item 10: Invents fantasy worlds or games and acts them out.
4. Item 24: Prefers to interact with others in an adopted role, or communicate through props/toys.

Total Score = (Items 6 + 8 + 10 + 24)/4

Reliability Analysis

	Cronbach's alpha
Controls	α=.700
ASC	α=.822
Overall	α=.837

Overall Inter-Item Correlation Matrix

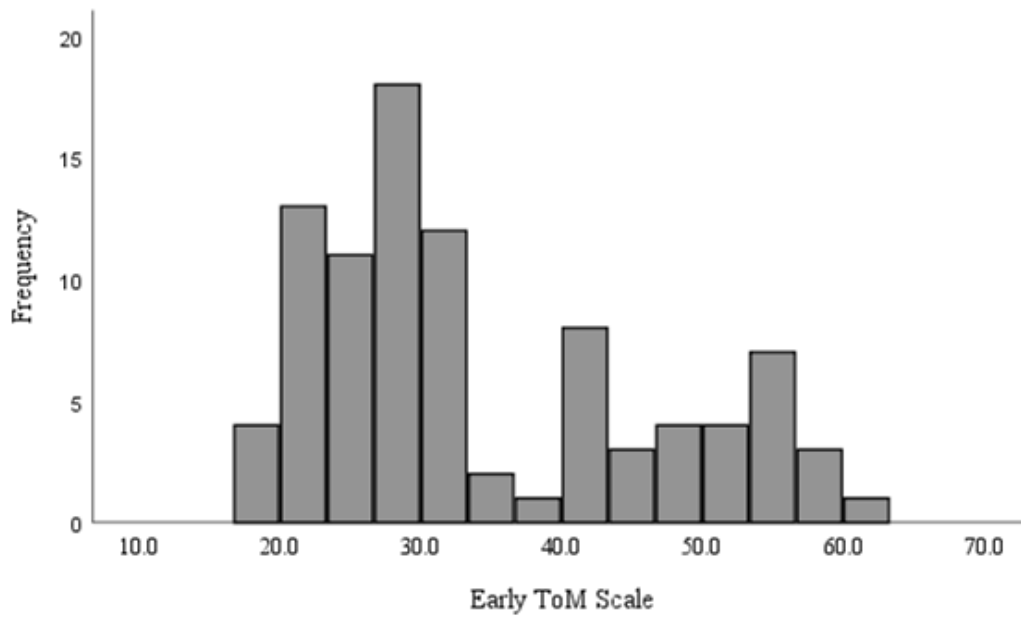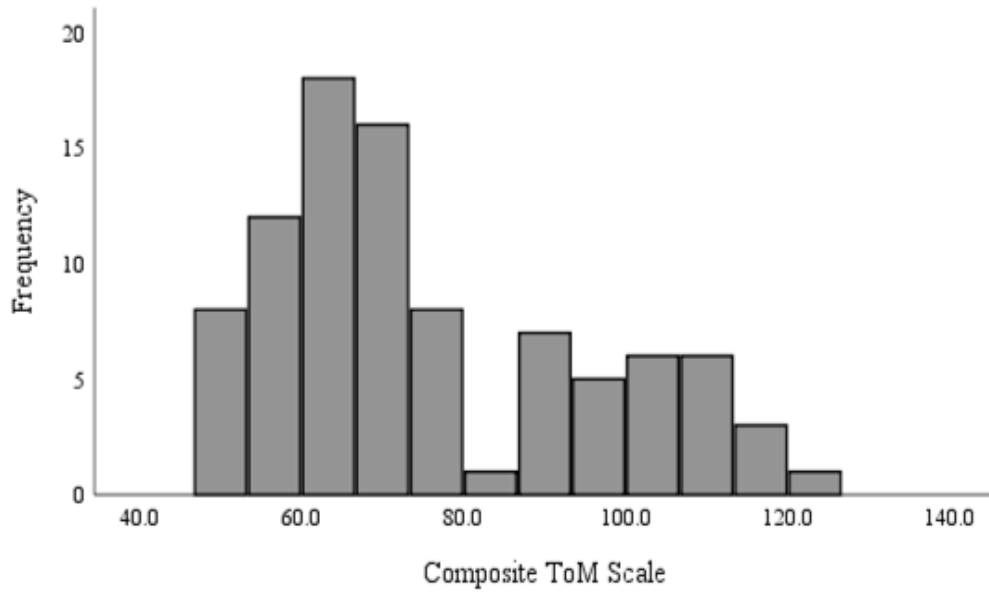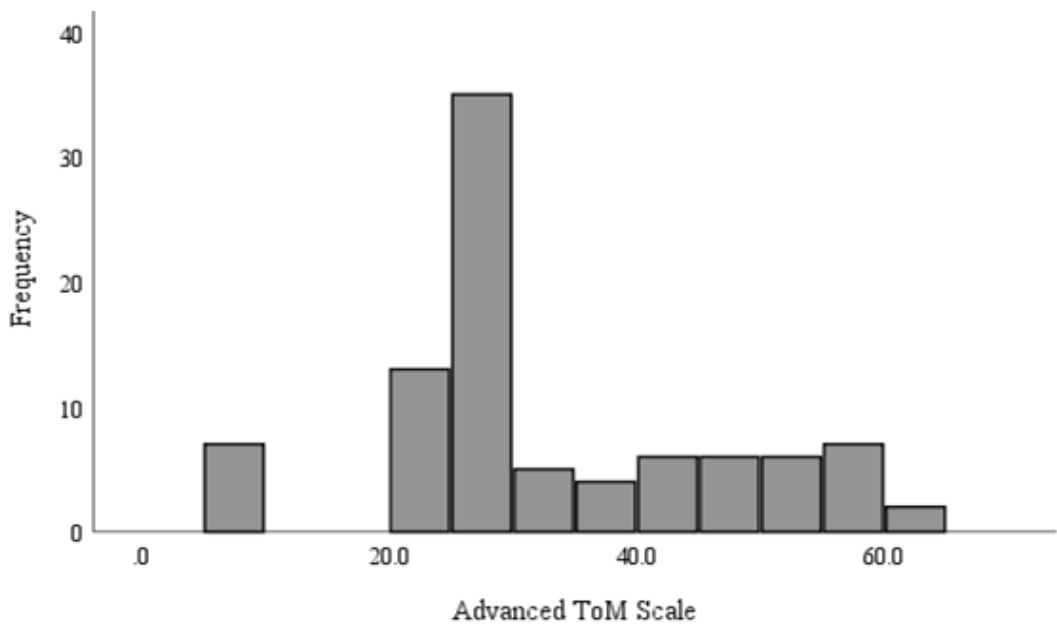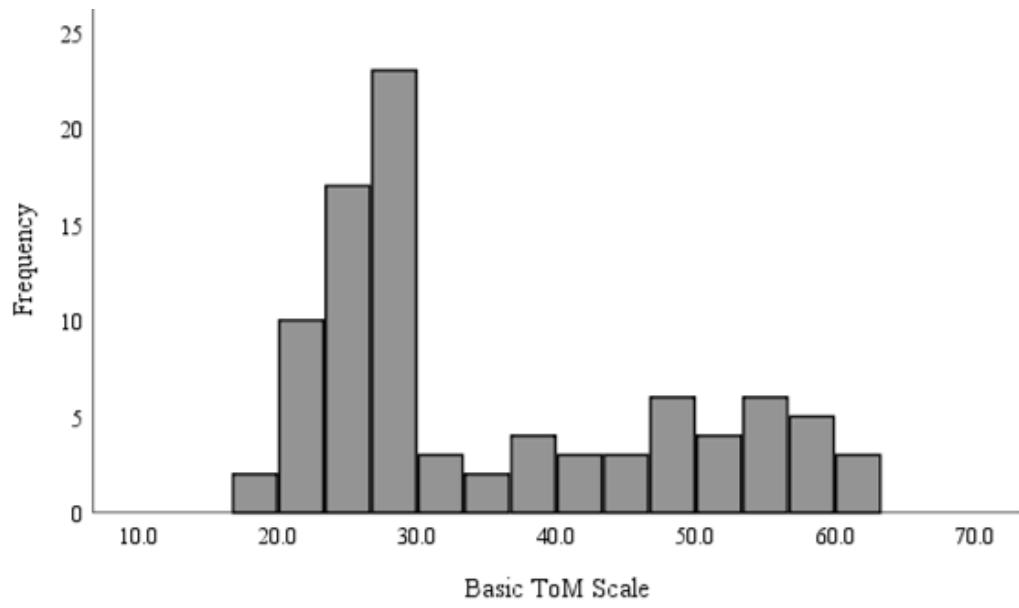|	Item 6	Item 8	Item 10	Item 24
Item 6		.552	.401	.614
Item 8			.676	.639
Item 10				.474
Item 24				

Overall Item Total Statistics

	Scale M if item deleted	Scale variance if item deleted	Corrected item-total correlation	Squared multiple correlation	Cronbach's Alpha if item deleted
Item 6	3.19	8.179	.612	.420	.817
Item 8	3.62	7.102	.763	.607	.748
Item 10	3.49	8.276	.607	.460	.819
Item 24	3.96	7.680	.692	.508	.782

Histograms to illustrate distributions of ToMI-2 data

Overall sample

Basic ToM Scale

Advanced ToM Scale

ASC Sample