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Systematic review documents limited empirical support for the practical application of the Theory of Mind model of ASD¹

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(1) Is it possible to teach Theory of Mind (ToM) and related skills to people with autism spectrum disorder (ASD)?

(2) Is there evidence to support the ToM model of ASD?

METHODS

Design: Systematic review and meta-analysis.

Data sources:Thirteen electronic databases and two trial registers were used to conduct a systematic search in July 2010, July 2012 and August 2013. These included the Cochrane Central Register of Controlled Trials; Ovid; Meline(R); Embase; CINAHLPlus; PsychINFO; ERIC; Social Sciences Index and Abstracts; ASSIA; Social Services Abstracts;

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metaRegister of Controlled Trials; ICTRP; UKCRN – UK Clinical Trials Network; ClinicalTrials.gov; and, Autism Data. The search strategy employed two concepts: the condition (ASD) and a search filter for ‘Randomised Control Trials’. No date or language restrictions were set; however, one article in French was excluded as it was awaiting translation at the time of publication.

Additional search strategies included contacting key authors directly for relevant published, unpublished or in-progress work, searching bibliographies of key articles for citations, and looking through conference proceedings from the *International Meeting for Autism Research*. Websites of specific journals known to regularly publish work on ToM were also searched.

Study selection and assessment: For inclusion in the review, studies had to empirically evaluate interventions that explicitly targeted ToM skills and/or were linked to ToM in one of four clearly defined ways: (a) explicitly state that the intervention was designed to teach ToM; (b) explicitly state that the intervention was designed to teach precursor skills of ToM including joint attention, imitation, and/or emotion recognition; (c) explicitly state that the intervention was based on or inspired by a ToM model of autism; or (d) explicitly state that the aim of the intervention was to test the ToM model of autism.

Interventions were excluded if they (a) did not meet any of the four criteria defined above; (b) were medical interventions; (c) targeted a particular behaviour rather than a cognitive skill; (d) were language-focused interventions; or (e) had a ‘broad-base’ with regard to methods and targets. The ToM interventions included in the review were compared with (a) treatment as usual/wait-list control, (b) ‘placebo’ interventions, and (c) intervention with no therapeutic content.

Articles were limited to randomized and quasi-randomized control trials with single-case experimental designs excluded. Participants of all ages were included if they had a

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confirmed diagnosis of an ASD (ICD-10, DSM-IV or DSM-5); participants with co-morbidities were also included.

After initial screening by one author, abstracts were reviewed by two of the authors independently. Inter-rater agreement was not reported. Authors state discrepancies were resolved with a third rater.

Two authors independently extracted data using a specifically designed data extraction form, discrepancies were resolved with a third author. Inter-observer agreement was not reported. Studies were coded in terms of methods (dose and frequency of intervention); diagnostic description of participant; type of intervention, including target, intensity, and duration; method of application (parent-mediated, therapist, school-based); risk of bias (sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcome assessments, incomplete outcome data, selective outcome reporting, and other sources of bias), and results (primary and secondary outcomes).

Outcomes:

MAIN RESULTS

The search yielded 22 studies for review, consisting of studies that explicitly taught ToM ($n = 5$) or precursor skills ($n = 17$). Precursor skills included emotion recognition ($n = 7$), joint attention and social communication ($n = 9$), and imitation skills ($n = 1$). Primary outcomes were 'Communication' (overall level of non-echoed language; stereotyped or idiosyncratic use of words or phrases; pointing; gestures; conversation), and 'Social function' (unusual eye contact; facial expressions directed to others; spontaneous initiation of joint attention; shared enjoyment in interaction; quality of rapport). Various measures were used to measure these outcomes including the Autism Diagnostic Observation Schedule (ADOS: Lord et al., 2000), Social Communication Questionnaire (SCQ: Rutter, Bailey, & Lord, 2003), Mullen Scales of Early Learning (Mullen, 1995), and Vineland Adaptive Behavior

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Scales (Sparrow, Balla, & Cichetti, 1984). Secondary outcomes were (a) intervention-specific and included change in targeted cognitive skills including emotion recognition, ToM (e.g., false belief, Happé's strange stories, faux-pas recognition), imitation, and play; (b) direct observation of participant behaviour or quality of interpersonal interaction, or both; (c) adult report of participant behaviour and skills or deficits; (d) participant cognitive skill, measured by standardised assessment; (e) acceptability of intervention; (f) rate of drop out; (g) economic data; and (f) follow-up effects. No outcome metrics (e.g., effect sizes) were calculated for individual outcomes, however these were calculated where possible for meta-analyses.

Three studies investigated the use of a computer program to deliver intervention (Bölte et al., 2002; Golan & Baron-Cohen, 2006; Hopkins et al., 2011), with emotion recognition as the target skill. Three studies examined the effect of a set of specially-designed cartoons on emotion recognition (Golan et al., 2010; Williams, Gray, & Tonge, 2012; Young & Posselt, 2012). Nine studies investigated the effects of one-to-one therapist-led interventions (Fisher & Happé, 2005; Goods, Ishijima, Chang, & Kasari, 2013; Hadwin, Baron-Cohen, Howlin, & Hill, 1996; Ingersoll, 2012; Kaale, Smith, & Sponheim, 2012; Kasari, Freeman, & Paparella, 2006; Landa, Holman, O'Neill, & Stuart, 2011; Ryan & Charragáin, 2010; Wong & Kwan, 2010), three used a therapist-led approach in a group treatment setting (Baghdadli et al., 2013; Begeer et al., 2011; Solomon, Goodlin-Jones, & Anders, 2004), and one study investigated a group music therapy approach (Kim, Wigram, & Gold, 2009). Four studies reported on an intervention which included a parent-training element (Begeer et al., 2011; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Schertz, Odom, Baggett, & Sideris, 2013; Solomon et al., 2004) and one study reported on a teacher-training intervention delivered within the classroom (Wong, 2013).

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Due to differing outcome variables and methods across studies, a meta-analysis for outcomes was only possible for three groups of studies: ‘social function’ (joint attention) studies, ‘emotion recognition’ studies, and ‘quality of interpersonal interaction/joint engagement’ (joint attention and social communication) studies.

The meta-analysis for ‘social function’ that included three studies (Goods et al., 2013; Ingersoll, 2012; Kaale et al., 2012) found no significant intervention effect on social behaviour using the Early Social Communication Scales (ESCS) (Cohen’s $d = 0.23$). One study not included in the meta-analysis found large gains in ‘showing’ and ‘responding to joint attention’ as measured using the ESCS (Kasari et al., 2006). Wong and Kwan (2010) found intervention effects on items of the ADOS (Lord et al., 2000) relevant to intervention content, but not on items unrelated to the intervention. Young and Posselt (2012) found no significant effects using a single item on the SCQ (Rutter, Bailey, & Lord, 2003) (‘Social Peer Interest’), despite the intervention having a positive impact on emotion recognition skills. Using standardised measures of life skills, Williams et al. (2012) found no intervention effects on social abilities. Thus overall, some support was found for teaching social function behaviors, albeit predominantly limited to the taught skill with minimal generalization.

The meta-analysis for ‘emotion recognition’ included four studies (Golan & Baron-Cohen, 2006; Hopkins et al., 2011; Ryan & Charragáin, 2010; Solomon et al., 2004). Focusing on the ‘older participant group’ results revealed a positive intervention effect on emotion recognition (Cohen’s $d = 0.75$). Included studies reported significant group differences in outcomes immediately post-treatment, as measured by recognition of facial emotion from static images (Golan & Baron-Cohen, 2006; Hopkins et al., 2011; Ryan & Charragáin, 2010; Solomon et al., 2004). Individual studies for emotion recognition excluded from the meta-analysis (Baghdadli et al., 2013; Williams et al., 2012; Young & Posselt,

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2012) that included measures to capture related constructs, found no generalization of the target skill to novel settings or stimuli.

Finally, the meta-analysis for ‘quality of interpersonal interaction/joint engagement’ behaviors as measured using direct observation of adult-child interactions, included two studies (Kaale et al., 2012; Kasari et al., 2010). Results indicated a positive intervention effect (Cohen’s $d = 0.55$) on joint engagement and transfer of therapist-taught social communication skills to a parent-child interaction when using novel assessments, but there was no generalization to new settings.

Due to the use of different outcome measures, 14 of the 22 studies were excluded from the meta-analyses. One study (Kaale et al., 2012) was included in two meta-analyses. Wong and Kwan (2010) and Young and Posselt (2012) used diagnostic measures to evaluate change in *communication*. Wong and Kwan (2010) documented gains in vocalizations directed to others, gestures and pointing using the ADOS (Lord et al., 2000) in the intervention group only; however, there were no between-group comparisons. Young and Posselt (2012) used the SCQ (Rutter et al., 2003) to examine change in eye contact and gaze aversion and found no intervention effects. Hadwin et al. (1997) examined the impact of ToM intervention on complex language skills, and found no effect of intervention on conversational skills.

Begeer et al. (2011), Fisher and Happé (2005), Hadwin et al. (1996) and Solomon et al. (2004) evaluated ToM using *explicit ToM* assessment. All studies reported positive outcomes of progress within, or close to, the taught context (such as the specific ToM task), but skills were not generalized to novel or more complex scenarios or skills. Two studies demonstrated gains in *imitation* skills as their primary outcome in interventions with toddlers (Ingersoll, 2012; Landa et al., 2011). Four studies included an assessment of *play* as a secondary outcome (Goods et al., 2013; Hadwin et al., 1996; Wong, 2013; Wong & Kwan,

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2010) and found mixed results. Goods et al. (2013) reported a positive intervention effect as measured using a 'Structured Play Assessment' of play *types*, but Wong (2013) reported no positive effects using the same measure to capture play *level*. Hadwin et al (1996) found no effect of teaching ToM understanding on symbolic play skills.

Seven studies documented improvements in *social skills* as a result of intervention (Begeer et al., 2011; Fisher & Happé, 2005; Hopkins et al., 2011; Ingersoll, 2012; Kim et al., 2009; Solomon et al., 2004; Wong & Kwan, 2010). Three studies produced conflicting results regarding effects of intervention using measures of language and general cognitive or adaptive ability (Kasari et al., 2006 reported in Kasari, Paparella, Freeman, & Jahromi, 2008; Landa et al., 2011; Schertz et al., 2013). Kasari et al. (2008) and Schertz et al. (2013) both reported significant gains in expressive language in the intervention group compared to the control, but Landa et al. (2011) reported no such difference; further evaluation provided no evidence that treatment continued to impact on language and cognitive outcome five years from baseline (Kasari, Amanda, Stephanny, Paparella, & Hellemann, 2012). Rates of drop out were reportedly low, with half of the included studies retaining the full sample for the study duration.

Only six studies included evaluation of *follow-up effects* after the immediate post intervention period had ended (Fisher & Happé, 2005; Hadwin et al., 1996; Kasari et al., 2006; Kasari et al., 2010; Landa et al., 2011; Ryan & Charragáin, 2010). These studies demonstrated maintenance of treatment gains in a range of measured skills, including ToM, social engagement, and emotion recognition, with some even demonstrating increases in functional play acts (Kasari et al., 2010) and ToM performance (Fisher & Happé, 2005) during the post-intervention period (Kasari et al., 2006). Only one study found no long-term impact of intervention on language and cognitive outcomes five years post-baseline (Kasari et al., 2012).

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AUTHOR'S CONCLUSIONS

The authors concluded that ToM and related precursor skills can be taught to individuals with ASD, but there is insufficient evidence documenting maintenance and/or generalization of these skills to novel contexts, or that teaching ToM has an impact on developmentally-linked abilities. Studies varied in quality with regard to blinding of participants and personnel, and under-reporting of other relevant practices such as sequence generation and allocation concealment. The authors also comment on the small sample sizes employed in the included studies, which ranged from ten (Bölte et al., 2002; Kim et al., 2009) to a maximum of 61 (Kaale et al., 2012). The authors were unable to conduct ASD subgroup analyses on the studies included in this review due to the broad range of participant ages and abilities, but suggest it is likely that differences would be found between specific diagnostic categories, age groups, and intervention delivery models.

Therefore, the authors highlight the need for multi-site systematic research linking intervention targets to individual characteristics, such as participant age and ability. This information is necessary to support decision making regarding treatment targets and starting points on an individual basis. The authors recommend that future research employ longitudinal methods to evaluate specific outcomes associated with subsequent abilities. There is a need for better outcome measures particularly with regard to ToM interventions. These measures should capture symptom severity in core diagnostic domains and studies should report primary outcome measures *a priori*. Secondary measures should have a clear rationale for inclusions, and where possible, should link to intervention components. Finally, the authors state that while there is some evidence that ToM, or related precursors skills, can be taught to individuals with ASD, the quantity and quality evidence for ToM and ToM-linked interventions, is inadequate to evaluate the ToM model of ASD.

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Commentary

Since Baron-Cohen, Leslie, and Frith's seminal paper in 1985, there has been considerable interest in the ToM model of ASD and its implications for remediation of associated impairments, particularly in the area of social communication. The main premise of this model is that impairments in the ability to understand internal mental states (beliefs, desires, knowledge) and their impact on behavior is a core impairment in ASD and explains the characteristic social and communicative challenges. Consistent with this, impairments in ToM have been well documented in individuals with ASD in the literature (see Baron-Cohen, 2001, for a review). However, the practical application of the ToM model is less clear and makes this systematic review timely. Interventions derived from the ToM model appear to have gained popularity over the past two decades and professionals urgently need accurate information about the efficacy of these interventions. The current review helps to meet this need while highlighting current gaps in knowledge in relation to empirically-supported treatments associated with the ToM model.

We found the review procedures to be well documented and soundly implemented with authors using a published protocol (Fletcher-Watson & McConachie, 2010), independent raters, and establishing clear inclusion/exclusion criteria. Minor deviations from the published protocol including additional acceptance of DSM-5 criteria (American Psychiatric Association., 2013) for diagnosis and removal of repetitive behaviours as an outcome measure consistent with recent ToM model conceptualizations, were outlined in the review. We focus the remainder of our commentary on three aspects of the review that warrant further discussion: types of interventions, types of outcome measures, and types of studies included.

Interventions eligible for inclusion in this review had to meet one of four criteria, one of which stated that interventions must "explicitly state that they are designed to teach

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precursor skills of ToM” (p10). Of the 22 papers included in the review, 17 targeted only ‘precursor skills’ including joint attention, imitation, and/or emotion recognition. Of these 17 studies, we found 10 made no explicit mention of ToM in relation to their intervention. This raises questions as to ‘if’ and ‘how’ these studies might address the ToM cognitive model and assumes the targeted skills (joint attention, imitation, and emotion recognition) are in fact precursors to ToM as theorized in the model. There is some controversy in the field as to whether these skills actually represent a more general social-cognitive competence rather than ToM-specific precursors (e.g., see Charman et al., 2000, for a discussion). Research is therefore needed to establish the nature of the relationship between these important developmental skills and the development of ToM. Inclusion of studies in the current review that target these skills could be questionable unless they specifically include a measure of ToM in order to investigate if changes in precursor skills impact on social/communication skills via changes in ToM. Inspection of the included precursor skills studies reveals that only one (Williams, 2012) included a measure of ToM and found training in emotion skills did not lead to changes in ToM.

The review authors come to an important conclusion that improved outcome measures, particularly in relation to ToM are needed and that existing measures are limited. Within these limitations, another consideration in relation to the included studies was whether assessment of ToM was a primary or secondary outcome. For the purpose of this review, the authors identified primary outcomes as measures of communication and social functioning. Direct assessment of ToM was identified as a secondary outcome. In order to test the value of the ToM model (the objective of this review), however, it would be beneficial to identify studies where ToM was a primary outcome and social/communication skills a secondary outcome. This would add to the evidence base of whether changes in ToM affect social/communication skills as hypothesized in the ToM model. Only five of the 22 included

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studies (Begeer et al., 2011; Fisher & Happé, 2005; Hadwin et al., 1996; Solomon et al., 2004; Williams et al., 2012) directly measured ToM skills. These studies generally found changes in only the taught skill with little or no generalization.

Thus the question remains whether training in ToM or its hypothesized precursors leads to changes in social and communicative domains as broadly measured in the included studies by tools like the Vineland Adaptive Behaviour Scales. Correlational studies examining this relationship have yielded mixed results (e.g., Fombonne, Siddons, Achard, & Frith, 1994; Frith, Happé, & Siddons, 1994). Further, some authors have argued and demonstrated that ToM may impact on only those social skills that require social maturity and/or perspective taking (e.g., Peterson, Slaughter, & Paynter, 2007). Consistent with this, a randomized-control trial published since this review found improvements in ToM were related to parent-reported ToM-linked social behavior, but not general social behavior (Begeer et al., 2015). In sum, examining changes in social and communication domains without including measures of change in ToM skills limits the conclusions that can be drawn from this review about the ToM model.

The final area for discussion in this commentary relates to the types of studies selected. The review included randomized and quasi-randomized trials and this resulted in only a small number of studies that directly assessed whether ToM could be taught (4 studies) or included ToM as an outcome measure (5 studies). Although randomized control trials are desirable because they provide a high level of evidence, Cochrane guidelines suggest inclusion of non-randomized studies, for example single-case experimental designs, as an option in these situations (Higgins & Green, 2011).

In sum, this review was timely and beneficial in identifying the current evidence base related to ToM applications. Evidence for ToM interventions and for the ToM model is limited and caution should be used when considering the use of ToM interventions with the

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aim of remediating social and communication skills in ASD. Four studies that directly assessed teaching ToM skills provide initial support for teaching ToM skills, but these skills failed to generalize beyond the immediate environment. The studies of ‘precursor’ skills suggest these are amenable to change (albeit once more limited in generalization) but fail to test if this leads to changes in ToM and subsequently in social and/or communication skills as would be predicted by the ToM model. There is a clear need for more research that directly tests whether interventions influence ToM skills, and whether (and under what conditions) this leads to improvements in associated social and communication skills. Such research will be facilitated by the development of better outcome measures.

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