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That's what I like: The use of circumscribed interests within interventions for individuals with autism spectrum disorder. A systematic review

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ABSTRACT

Background: Circumscribed interests (CI) are a subcategory of restricted and repetitive behaviors that occur commonly in individuals with autism spectrum disorder (ASD). CI are characterized by an intense and focused interest in a narrow range of subjects. The purpose of this systematic review is to determine how interests are incorporated within interventions for individuals with ASD across the lifespan; what symptoms, domains and outcomes these interventions target; and the effectiveness of such interventions.

Method: The methods used within this review were consistent with those recommended by the Cochrane and Campbell Collaborations. Inclusion criteria were based on three predetermined categories: (1) Study Population; (2) Intervention Design; and (3) Outcome Variables. Data were extracted and coded based on these three predetermined categories.

Results: 246 full-text articles were assessed for eligibility, of which 31 studies were eligible for data extraction. The majority of studies were single subject designs (k = 28) and focused on toddlers/preschool (k = 13) or school-aged children (k = 17). Common interests utilized were TV shows or movies (N = 21), popular characters (N = 18), computers/video games (N = 12) and transportation (N = 11).

Conclusions: Results suggest that the inclusion of CI within interventions can lead to positive effects across a number of domains. More research is required to examine the effects of individualized interests within group design studies. Methods for this review were registered with PROSPERO (42016036981).

1. Introduction

Circumscribed and restricted interests are a subcategory of restricted and repetitive behaviors (RRBs) that occur commonly in individuals with autism spectrum disorder (ASD). Compared to both the domain of social-communication and other behaviors classified under the umbrella of RRBs, considerably less is known about circumscribed interests (CI; also referred to in the literature as intense, restricted, perseverative, preferred or special interests).

Circumscribed interests are characterized by an intense and focused interest in a narrow range of subjects. Individuals with ASD often organize their activities rigidly around these interests (Klin, Danovitch, Merz, & Volkmar, 2007; South, Ozonoff, & McMahon, 2005). Some CI do include topics, interests and objects that overlap with those observed in typical development (DeLoache, Simcock,

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& Macari, 2007). Examples of common CI that are also observed in typical development, particularly typically developing boys, include Legos, computers and automotives (South et al., 2005). However, other examples are often idiosyncratic, less functional and less age-appropriate, such as an interest in washing machines or watches and clocks (Mercier, Mottron, & Belleville, 2000; South et al., 2005).

Reports suggest that between 75% and 95% of individuals with ASD have at least one CI (Klin et al., 2007; Turner-Brown, Lam, Holtzclaw, Dichter, & Bodfish, 2011), with a high proportion of individuals having multiple (Turner-Brown et al., 2011). Research indicates that unlike other types of RRBs (such as repetitive motor actions), the impact and intensity of CI does not always improve with age (South et al., 2005). Further, earlier development of CI has been found to be predictive of more extensive and limiting RRBs overall in later childhood (Troyb et al., 2016).

Compared to hobbies and interests in typical development, CI in ASD have been found to have a profound effect on learning and adaptive behavior (Varni, Lovaas, Koegel, & Everett, 1979) as well as socialization opportunities (Boyd, Conroy, Mancil, Nakao, & Alter, 2007; South et al., 2005). Parents frequently rate CI in ASD as more intense and as more likely to interfere with everyday activities than interests reported in typical development (Cascio et al., 2014; Klin et al., 2007; Turner-Brown et al., 2011). Individuals also recognize the potential negative impact of their CI (Mercier et al., 2000). For example, CI in ASD may be accompanied by more impairment and their interests are less likely to involve other people (Anthony et al., 2013; Turner-Brown et al., 2011).

The effects of CI on development extend to both an attentional and neural level. A number of eye tracking studies have found that when social images are presented at the same time as images of commonly occurring CI, children with ASD fixate more on CI images and are slower to fixate on social images (Sasson & Touchstone, 2014; Sasson, Turner-Brown, Holtzclaw, Lam, & Bodfish, 2008; Unruh et al., 2016). At a neurobiological level, both images of commonly occurring CI and an individual's CI have been linked to increased activation in systems associated with affective circuits (Cascio et al., 2014; Sabatino et al., 2013), sustained attention (Cuthbert, Schupp, Bradley, Birbaumer, & Lang, 2000) and motivation (Keil et al., 2002). Taken together, these studies indicate that attention to social stimuli and cognition may be adversely affected by the presence of CI.

While there are a number of potentially negative consequences of CI, there are also some potential benefits. For example, they may represent *islands of ability* for individuals and a source of great pleasure and shared enjoyment (Mercier et al., 2000). As such, there seem to be positive aspects that accompany CI. A number of studies have indicated the increased reward value of CI for individuals with ASD. Sasson, Dichter, and Bodfish, (2012) reported that adults with ASD rated images of commonly occurring CI as more pleasant than non-CI related images. In a series of semi-structured interviews with adults with ASD, Mercier et al. (2000) found that CI provided them a sense of well-being, personal validation, and were seen as islands of ability for some individuals. This expertise associated with CI also has been observed at the neurobiological level. At a neural level, both Grelotti et al. (2005) and Foss-Feig et al. (2016) reported more robust activation of the Fusiform Face Area (FFA) in individuals with ASD compared to TD controls when viewing images of their own, personalized interests suggesting enhanced visual expertise related to their personalized interests.

Due to the increased motivation surrounding CI, incorporating them into interventions may lead to improved outcomes for individuals with ASD. Thus, it is assumed that individuals would naturally be more motivated to engage and participate in interventions that use high interest items (Grove, Hoekstra, Wierda, & Begeer, 2018). Previous research indicates that using CI as reinforcement may be more effective than alternative reinforcers such as food (Charlop, Kurtz, & Casey, 1990), and that integrating CI into an intervention model or an intervention activity may be beneficial for individuals with ASD (e.g. Baker, Koegel, & Koegel, 1998). For example, power cards (Gagnon, 2001) are an existing intervention approach designed around an individual's CI, and have led to improvements in adaptive behavior for individuals with ASD (e.g. Davis et al., 2010). Despite promising findings, interventions incorporating CI within the treatment model/delivery are not commonly reported, particularly in randomized-controlled trials with large sample sizes. As a result, it is important to further examine the ways in which CI are being used in interventions and the outcomes resulting from their use.

1.1. Aims

Circumscribed interests represent an understudied area of ASD; however, they are considered clinically relevant and potentially important for treatment. Despite this, to our knowledge, there exist a limited number of intervention studies that incorporated an individual's CI as a vehicle for change. The purpose of this systematic review is to determine how CI are incorporated within interventions for individuals with ASD; what symptoms, domains and outcomes these interventions target; and the effectiveness of the interventions. This review differs from previous reviews on a similar topic (e.g. Dunst, Trivette, & Hamby, 2012) by specifically focusing on CI used within treatment delivery rather than more global preferred objects, such as candy or those selected via a preference assessment, or topics/objects assumed to be common interests in ASD (such as Legos and robots) but not confirmed to be a CI. The overarching goal is to synthesize the intervention research and utilize the findings to provide recommendations for research and practice.

2. Method

The methods used within this review were consistent with those recommended by the Cochrane and Campbell Collaborations (Noonan & Bjørndal, 2010) and reported according to the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA; Moher, Liberati, Tetzlaff, Altman, & The PRISMA Group, 2009). The methods for this systematic review are registered with PROSPERO (ID: 42016036981) and can be accessed at http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID = CRD42016036981.

2.1. Selection criteria

Inclusion criteria were based on three predetermined categories: (1) Study Population; (2) Intervention Design; and (3) Outcome Variables.

2.1.1. Study population

The study must have reported the child had a diagnosis or label consistent with ASD. This criterion could have been meet in the following ways: Participant had an educational label of ASD (PDD-NOS, Asperger's, Autism), community clinical diagnosis of ASD, and/or a validated ASD screening or diagnostic tool was used to confirm the child was on the autism spectrum. If studies included multiple subjects with different clinical diagnoses, then if possible, only the subject with ASD was included in data extraction. No exclusions were made based on age, race / ethnicity or sex. We excluded studies that included subjects with a diagnosis of a comorbid genetic syndrome (such as Fragile X) or if the primary diagnosis that was not ASD.

2.1.2. Intervention design

Inclusion criteria focused on interventions using CI as a mode of intervention delivery and/or as reinforcers within the intervention. For the purpose of this study, CI were defined based on DSM-5 criteria;

> "Highly restricted, fixated interest that are abnormal in intensity or focus (e.g. strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interests). (American Psychiatric Association, 2013).

Interests did not need to be *unusual* to be included in the review. *Typical* interests that were defined as pervasive or circumscribed by the study authors were also included. Unlike some reviews of interest-based learning interventions that precede this one (e.g. Dunst et al., 2012), we have focused solely on *established individualized CI*. As such, the study must have reported that the interests being used were indeed the child's individualized CI and not simply common interests found in ASD (e.g. robots, Legos) not confirmed as individualized to study participants. Arbitrary reinforcers (such as food) were not considered CI. Circumscribed interests had to be confirmed prior to intervention onset or be an inclusion criterion for the study (e.g. researchers required participants to have a specific intense CI to be included). Experimental, quasi-experimental and single subject design (SSD) studies were included. Unlike previous reviews (e.g. Ninci, Rispoli, Burke, & Neely, 2018), we did not require that studies have a comparison condition (e.g. interest-based vs. low preference) for inclusion. As we anticipated that the majority of our studies would be SSD studies, the requirement of a control comparison condition would have severely reduced the number of eligible studies. Further, within the context of SSD methodology, the individual serves as his / her own control. We did not exclude for dosage, frequency or method of administration. We excluded grey literature and descriptive case studies and studies using common reinforcers, such as food, instead of personalized interests. Articles not published in English were excluded.

2.1.3. Outcome variables

All studies had to include at least one primary outcome directly related to participant behaviors. Social, adaptive, cognitive, work-based, communication, behavioral and academic outcomes were all included. We did not include studies if the primary outcome was not directly related to the participant, such as caregiver stress and social validity of intervention.

2.2. Search methods

A trained health sciences librarian (STW) performed a comprehensive electronic search of publications using the following databases: PubMed, Web of Science Core Collection, Embase via Elsevier, Cochrane Central Register of Controlled Trials (CENTRAL), PsycInfo via EBSCO, Google Scholar, ERIC via EBSCO, and CINAHL via EBSCO. We also hand searched specific journals relevant to the subject area for additional resources (e.g. Journal of Autism and Developmental Disorders, Autism Journal, Autism Research, Journal of Developmental Disabilities, Focus on Autism and Other Developmental Disabilities, Research in Autism Spectrum Disorders). Hand searching allowed for a more thorough review of journals specific to ASD for any publications that were not indexed when the journal was indexed. All journals that were hand-searched were available through the databases referenced previously.

All database results were collected from the inception of the database through June 2016. Search updates were conducted prior to submission to ensure all relevant records were located. Search terms were used to retrieve articles addressing the two main concepts of the search strategy: (1) circumscribed interests; and (2) autism or autistic disorder. The exact search strategies used in each of the electronic databases are reported in Appendix A. Because electronic search strategies may be incomplete, we also manually searched several journals' Table of Contents (see previous material and Appendix A) to ensure a comprehensive review of the literature. Results from the database searches were downloaded to EndNote and deduplicated. Then, all references were uploaded to Covidence Systematic Review software (https://www.covidence.org), a web-based tool designed to facilitate and track each step of the abstraction and review process.

Two reviewers (CH and JA) independently screened the titles and abstracts for all located studies in Covidence. Conference abstracts, non-peer reviewed articles and Masters Theses were excluded. However, if a Masters thesis resulted in peer-reviewed publications, the peer-reviewed publication was included. Doctoral dissertations were included. If doctoral dissertations also resulted in a peer-reviewed publication, we only reviewed any additional variables in the dissertation that were not included in the peer-reviewed publication so not to duplicate data included in this review. For example, if a dissertation reported five outcome variables and the corresponding peer-reviewed article included only two of these, we reported on the other three. If published articles or

dissertations included multiple studies, but not all met inclusion criteria for this review, we only included data and results from studies that met criteria.

After each stage, the reviewers met to discuss any studies where there was disagreement on inclusion or exclusion criteria. Any study that could not be resolved by the two initial reviewers, the full-text was sought and any remaining disagreements were decided by the senior author. The coders had 96.5% agreement at the title and abstract level. The majority of conflicts were resolved between the two coders. Forty conflicts were resolved by the senior author at this stage (0.28% of the total articles – 42 titles/abstracts). At the full-text stage, the two coders agreed on 86% of the articles. After discussion, 4.5% (11 articles) of the full-text article conflicts were resolved by the senior author.

2.3. Variable definitions, data extraction and coding

Data were extracted and coded based on the three predetermined categories: (1) Study sample and research design; (2) Intervention design; and (3) Outcome variables and results. The same two independent coders double coded all variables. Coders agreed on 92% of the total coded extracted variables. Coders met to discuss their coding and discrepancies were resolved through mediation.

2.3.1. Study sample and research design

Seven variables were coded for study sample and research design (Table 1). Four variables focused on participant characteristics: study N, age of subjects, diagnostic category and how diagnosis was confirmed/verified. Three variables focused on the research design: study design (Experimental, quasi-experimental and SSD), the participant's CI and how the research team confirmed it. Participants' CI also were grouped into categories (based on categories used in previous literature and generated through data extraction), however, categories were not mutually exclusive; therefore, a single CI could be assigned to multiple categories. For example, Thomas the Tank would be assigned to TV Characters, TV Shows and Transportation.

2.3.2. Intervention design

Three variables related to intervention characteristics were extracted and coded (Table 2); *Intervention Approach* (i.e. *how* the intervention was delivered. E.g. video modeling, social groups, power cards); *Delivery Method* (i.e. *who* delivered the intervention. E.g. parents, peers, researchers); and *Intervention Protocol* (a brief description of the intervention protocol).

2.3.3. Outcome variables and results

Three variables related to outcomes were coded. We categorized the outcome variable (e.g. joint attention, adaptive behavior), how the outcome was measured, and provided a brief summary of the results for each subject/group of subjects (Table 3). Results were rated as *positive, negative* or *mixed* based on the pooling of findings across variables and subjects. If the majority of subjects reported gains across outcome variables, this was rated as *positive.* If there were more negative results, this was rated as *negative.* If the study reported a balance of positive, negative or non-significant change, this was rated as *mixed.* Maintenance and/or generalization of results were rated in the same way.

Data were organized alphabetically in Tables 1 and 2. Outcome data (Table 3) is organized by intervention approach (*how* the intervention was delivered).

2.4. Quality assessment and risk of bias

The two independent coders (CH and JA) also assessed the study's risk of bias using the methodology and risk of bias tool detailed by Cochrane and the Campbell Collaboration (Noonan & Bjørndal, 2010) and developed by Reichow and colleagues (Barton, Reichow, Schnitz, Smith, & Sherlock, 2015, Reichow, Barton, & Maggin, 2016, Barton, Pustejovsky, Maggin, & Reichow, 2017). For SSD, we used the following domains to assess risk of bias: sequence generation, subject selection, blinding of subjects, fidelity, blinding of outcome assessor, dependent variable reliability and data sampling. For group designs, we used the following domains to assess risk of bias: sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcome assessment, incomplete outcome data, and selective outcome reporting. Discrepancies were resolved through consensus, and when consensus could not be reached, the senior author made the final judgement on the risk of bias. For the SSD, the two coders met consensus on 83.7% of the variables. For the group design studies, the coders agreed on 91.6% of the variables. These differences were resolved through discussion with the senior author who has extensive experience in SSD and group design as well as the manuscript's fourth author, who has extensive experience conducting systematic reviews.

A summary of the risk of bias scores are provided in Table 3. These scores represent the number of *low* ratings summed across items from a total of seven (SSD) or eight (group design) categories. Therefore, a higher score indicates less risk of bias. These data are also summarized in Figs. 4 and 5 across SSD and group studies, with green indicating a low risk of bias, yellow indicating unclear and red a high risk of bias. Risk of bias scores for each study are summarized in Figure 6 (Supplementary). A copy of the risk of bias tool is available from the authors upon request.

Table 1
Sample Characteristics.

Study	N	Age Range (Y.M)	Age Category	Diagnosis	Diagnosis Method/ Confirmation	Research Design	Circumscribed Interest	How Confirmed
Adams (1998)	5	3.1 – 4.11	Toddler and Preschool	Autistic Disorder or Asperger's Syndrome (DSM IV)	DSM-IV Criteria and CARS	Multiple baseline across tasks design	Letters, Jewelry/crystals, Thomas the Tank, & Musical instruments	Teacher & Parent Interviews
Angell et al. (2011)	1 (2 non ASD)	11		Autism	Diagnosed by outside agency	Single subject ABABAB withdrawal design	Computer games, electronics, & musical toys (yet Dad used on power cards)	Staff & family interviews
Ashbaugh et al. (2017)	3	19.2-24	Adulthood	Autist (DSM –IV-TR or DSM-5)	Diagnosed by outside agency and verified by team	Multiple baseline across participants design	Not reported	Subject and parent report
Baker (2000)	3	5.5 – 6.10	School Aged	Autism (DSM IV)	Diagnosed by outside agency and screened with CARS	Multiple baseline across participants design	Number lines on paper, cars, crashing cars, & vacuums	Teacher interview Child observations
Baker et al. (1998)	3	5.4 – 8.9	School Aged	Autism (DSM IV)	Diagnosed by outside agency	Multiple baseline across participants design	Disney	Teacher, aides and parent interviews Child observations
Boyd et al. (2007)	3	5.3-5.8	School Aged	Autism	Diagnosed by outside agency and screened with CARS	Alternating treatment design	Thomas the Tank & construction trucks	Teacher interview Preference assessment Choice condition with peers
Campbell and Tincani (2011)	3	6	School Aged	Autism & Pervasive Developmental Disorder-Not Otherwise Specified	Diagnosed by outside agency with CARS or GARS-2	Multiple baseline across participants design	Trains, Carla (character from the Starfall website), & Carl & Russel from Up (movie)	Teacher interview Participant interview Child observation
Carnett et al. (2014)	1	7	School Aged	Autism	Diagnosed by outside agency with CARS	Alternating treatment design	Jigsaw puzzles	Teacher interview Preference assessment
Davis et al. (2010)	3	16.3-17.8	Adolescence	Asperger's Syndrome (DSM IV-TR)	Diagnosed by outside agency	Multiple probe across participants design	Atlanta Braves, Yu-Gi-Oh Comic, & college basketball	Subject report
Dunst et al. (2010)	17	1.11 – 5.11	Toddler and Preschool	Autism	Diagnosed by outside agency	Group design (Quasi-experimental) – high vs. low interest groups	Not Reported	Parent Interview Investigator-administered interestingness scale
Dunst et al. (2011)	17	1.11 – 5.11	Toddler and Preschool	Autism	Diagnosed by outside agency	Group design (Quasi-experimental) – high vs. low interest groups	Not Reported	Parent Interview Investigator-administered interestingness scale
El Zein et al. (2016)	1	8	School Aged	Autism	Not Reported	Alternative treatment design	Cars	Four step assessment process: Child observation Teacher questionnaire Subject questionnaire Preference assessment
Haymes (1995)	3	6-10.9	School Aged	Autism (DSM III)	Not Reported	Multiple baseline across participants design	Wheel of fortune; Words, (writing/ spelling/meaning) Beauty and the Beast, Animals, Magic Copier game, Bart Simpson, Water play, & Sesame street, Mario Brothers	Not reported
Jung and Sainato (2015)	3	5.10 – 6.9	School Aged	PDD-NOS	Not Reported	Multiple probe across participants design	Princesses (Belle)	

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Table 1 (*continued*)

Study	N	Age Range (Y.M)	Age Category	Diagnosis	Diagnosis Method/ Confirmation	Research Design	Circumscribed Interest	How Confirmed
								Teacher interview Parent survey Child observations
Keeling et al. (2003)	1	10	School Aged	Autism (DSM IV)	Not Reported	Multiple baseline across conditions design	Power puff girls	Not reported
Koegel et al. (2012)	3	11 - 14	School Aged; Adolescence	Autism	Diagnosed by outside agency and verified by team according to DSV-IV criteria	Repeated measures multiple baseline across participants design	Movies	Not reported
Koegel et al. (2012)	3	9-12	School Aged	Autism	Diagnosed by outside agency	Noncurrent multiple baseline across participants design	Animated TV characters, board games, cooking, arts and crafts, & video games.	Preference assessment
Koegel et al. (2013)	7	14-16	Adolescence	Autism (DSM IV-TR)	Diagnosed by outside agency and verified by team according to DSV-IV-TR criteria	Multiple baseline across participants	Movies, ultimate frisbee, video games, basketball, computer graphics, and cooking	Subject interview
Koegel et al. (2013)	3	21-23	Adulthood	Asperger's Syndrome (DSM IV-TR)	Diagnosed by outside agency	Multiple baseline across participants	Music/dancing, math, computers, sports, exercise and fitness	Subject report
Koegel et al. (2017)	2	8-9.6	School Aged	Autism Spectrum Disorder	Diagnosed by outside agency	Alternating treatment design	Super Mario Brothers, Cars, trucks, & traffic signs	Observation, teacher, parent, & child report
Kryzak and Jones (2015)	3	2 - 8	Toddler and Preschool; School Aged	Autism	Diagnosed by outside agency Screened by 1st author	Multiple probe across participants design	Trains & letters	Teacher & parent report Child observations
Kryzak et al. (2013)	3	3 - 14	Toddler and Preschool; Adolescence	Autism	Not reported	Multiple probe across participants design	Trains	Parent report Preference assessment
Kuligowski (2010)	5	9-12	School Aged	P1, 2 & 4 PDD-NOS P3 Autistic Disorder P5 Asperger's Syndrome	Not reported	Single subject AB design	Mets baseball team, iCarly (TV show), SpongeBob, sports, & Charles Darwin	Social skills leader report
Lepper et al. (2017)	2	11-12	School Aged	Autism	Diagnosed by outside agency; CARS completed (unclear if by study team or outside agency	Alternating treatment design	Specific toys, cartoons, video games, statements about time and time concepts, fantasy world, fairy tales/ movie, video game & cartoon character	Parent report Observations during functional analysis
MacCormack (2016)	4	11-13	School Aged	Autism	Not reported	Pre-Post	Minecraft	Parent & Child report
Marshall (2017)	2	15-17	Adolescence	ASD	Not reported	Alternating treatment design	Cars (movie); Pokemon	Parent and teacher report Subject report Direct observations
Ohtake et al. (2015)	1	12	School Aged	Autism	Not reported	Multiple baseline across behaviors design	Initial D (Japanese carton)	Teacher report & questionnaire
Spencer et al. (2008)	1	5	School Aged	Autism	Not reported	AB design	Lightning McQueen	Not reported
Trivette and Dunst (2011)	17	1.11-5.11	Toddler and Preschool	Autism	Diagnosed by outside agency with CARS, ADOS or ABC	Group design (Quasi-experimental) – high vs. low interest groups	Details not available	Parent interview
Vismara (2005)	3	2-3	Toddler and Preschool	Autism		Phase reversal (ABA or BABA) design with	Letters & numbers	CI was inclusion criteria for study

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Table 1 (*continued*)

Study	N	Age Range (Y.M)	Age Category	Diagnosis	Diagnosis Method/ Confirmation	Research Design	Circumscribed Interest	How Confirmed
					Diagnosed by outside agency and verified by team according to DSV-IV criteria	alternating treatment design in final condition		Parent interview Informal child observations
Vismara and Lyons (2007)	3	2-3	Toddler and Preschool	Autism	Diagnosed by outside agency and verified by team according to DSV-IV criteria	Phase reversal (ABA or BABA) design with alternating treatment design in final condition	Letters & numbers	CI was inclusion criteria for study Parent interview Informal child observations

Table 2
Intervention Design.

Study	Outcome Variable	Intervention Approach (*how* the intervention was delivered)	Delivery Method (*who* delivered the intervention)	Brief Description of Intervention Protocol
Adams (1998)	Improving independent work behavior	Activity/task based (school)	Researchers	CI incorporated into individual child tasks at school
Angell et al. (2011)	Decreasing latency in response to cues for classroom transitions	Power cards	Classroom Staff	Used power cards to help with transitions in classroom
Ashbaugh et al. (2017)	Increasing social integration	Social groups (structured social planning)	Researchers and Peer mentors	Interventionist helped participant research events, clubs and activities related to CI. Interventionist also provided training on how to find activities and met once a week to plan activities. Peer mentors provided support and attended activities with participant.
Baker (2000)	Increasing social play interactions	Social groups (games with siblings)	Siblings	CI incorporated into bingo game with siblings
Baker et al. (1998)	Increasing social play interactions	Social groups (games with peers)	Adults and Peers	School games modified to center around each child's CI
Boyd et al. (2007)	Increasing social behavior toward peers	Social groups (play activities)	Peers	Peers were trained to offer CI or low preference item. CI imbedded into play with peers
Campbell & Tincani (2011)	Increasing direction following	Power cards	Classroom Staff	Power cards with CI used during free play
Carnett et al. (2014)	Decreasing challenging behavior & increasing on-task behavior	Activity/task based (literacy activity)	Researchers	Compared regular "penny" token economies with token economy centered around CI
Davis et al. (2010)	Increasing and improving conversational skills	Power cards	Teacher and Peers	CI based power cards used by teachers to promote conversations with peers
Dunst et al. (2010)	Increasing learning opportunities	Activity/task based (home)	Parents	Researchers interviewed parents to determine daily routines and activities to imbed CI into
Dunst et al. (2011)	Increasing developmental progress	Activity/task based (home)	Parents	Researchers interviewed parents to determine daily routines and activities to imbed CI into
El Zein et al. (2016)	Increasing reading skills & comprehension	Activity/task based (reading)	Researchers	CI imbedded into reading text
Haymes (1995)	Increasing social initiations	Video modelling	Researchers	CI included into some videos. Asked children to watch and then imitate with peer in playroom
Jung and Sainato (2015)	Increasing engagement with game & peers	Video modelling	Researchers and Peers	CI incorporated into a board game; Video model of two actors playing board game; Played game with peers
Keeling et al. (2003)	Increasing sportsmanship	Power cards	Researchers	CI integrated into a power card scenario/script of game
Koegel Vernon et al. (2012)	Increasing social interactions with peers	Social groups (social clubs)	Researchers and Peers	Participants attended social clubs formed around CI with peers
Koegel and Fredeen et al. (2012)	Increasing social interactions with peers	Social groups (social clubs)	Researchers and Peers	Participants attended social clubs formed around CI with peers
Koegel and Ashbaugh et al. (2013)	Increasing engagement and social initiations	Social groups (social clubs)	Researchers and Peers	Socialization opportunities that incorporated CI were implemented during lunchtime social clubs
Koegel and Kim et al. (2013)	Increasing social engagement	Social groups (structured social planning)	Researchers	Interventionist helped participants research community events and clubs related to CI
Koegel et al. (2017)	Increasing socialization	Activity/task based (recess)	Non-trained peers	CI embedded into recess activities announced prior to beginning recess; peers participated at will
Kryzak and Jones (2015)	Initiating joint attention (IJA)	Activity/task based (prompt fading and reinforcement at home)	Researchers, Teachers and Parents	Interventionists used prompt fading and reinforcement while engaging the child in CI related activities at home
Kryzak et al. (2013)	Responding to joint attention (RJA)	Activity/task based (home)	Researchers	RJA opportunities imbedded within CI activities at home
Kuligowski (2010)	Improving social skills	Power cards	Researchers	CI imbedded into power cards that were used in conjunction with social skills group activities
Lepper et al. (2017)	Increasing conversation topics	Social conversations	Researchers	Lag reinforcement contingencies embedded within CI-related and unrelated conversations
MacCormack (2016)	Increasing social competence	Social groups (play/video play)	Researchers, Volunteers, and Peers	Structured and free play with Minecraft and circle time with video social stories.

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Table 2 (*continued*)

Study	Outcome Variable	Intervention Approach (*how* the intervention was delivered)	Delivery Method (*who* delivered the intervention)	Brief Description of Intervention Protocol
Marshall (2017)	Increasing reading skills & comprehension	Activity/task based (reading)	Researchers	CI imbedded into reading text at varying frequencies
Ohtake et al. (2015)	Improving bathroom-related behaviors	Video modelling	Teachers	Video hero (CI) modelled four target bathroom behaviors
Spencer et al. (2008)	Increasing social, academic, & behavioral skills	Power cards	Teachers	Power cards used to teach sportsmanship skills with CI imbedded
Trivette and Dunst (2011)	Increasing social-affective behavior	Activity/task based (home)	Parents	Researchers interviewed parents to determine daily routines and activities to imbed CI into
Vismara (2005)	Increasing social sharing and initiating joint attention	Motivational pivotal response training (PRT)	Researchers and Parents	Motivational PRT used in conjunction with CI
Vismara and Lyons (2007)	Improving joint attention behaviors	Motivational PRT	Researchers and Parents	Motivational PRT used in conjunction with CI

CI = circumscribed interest.

Table 3
Outcome by Intervention Approach.

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
Power cards				
Angell et al. (2011) P1 only	Classroom behavior/transition	Observed mean transition latency time	1. P1 Transition latency decreased when using power-card & increased at baseline	4
Campbell and Tincani (2011)	Direction following (verbal directions by adult)	Observations coded during morning playtime Social validity survey completed by staff	1. All participants increased in direction following in both scenario & card conditions; P1 effects not maintained; Some maintenance for P3. Effects maintained for P2; P1 decreased to baseline; P2 maintained direction following 2. Staff reported intervention as feasible & acceptable	4
Davis et al. (2010)	Conversational skills (others-focused conversation) Social validity	Observational data sheet & stopwatch used to record time engaged in others-focused conversation Social validity rated by peers & parents	1. All participants increased others-focused conversation during intervention; Only P2 &P 3 generalized 2. Parents rated intervention as important & 2/3 wanted children to have more others focused conversations	2
Keeling et al. (2003)	Sportsmanship skills (whining, screaming, during gross motor, board game, & card game activity)	Amount of time whining & screaming recorded using a stopwatch	1. Gross motor whining decreased during intervention (no screaming during gross motor) 2. Board game whining & screaming decreased 3. Card game whining decreased (no screaming during card game 4. No generalization from 1 st to second but started to with 3rd game	0
Kuligowski (2010)	Child resiliency; Social skills; Observed target behaviors	Behavioral observations & standardized questionnaires (Parent report - Social Responsiveness Scale; Self-report – Resiliency Scales for Children & Adolescents; Sense of Mastery)	1. Target behaviors improved in 4 out of 5 participants (no data for P2). 1 large effect size (P4), 1 medium (P1) & 2 small (P3 & 5) 2. Frequency of engaging in conversation with others did not increase in P1 & P3 3. P2 did not show improvement in speaking off topic 4. P4 increased in their frequency of interrupting others 5. P5 showed increase in their tolerance for others 6. P1 optimism remained the same. P5 improved in their feelings of optimism 7. P3 & P4 improved in their self-reports of adaptability; P1 & P5 remained the same 8. Varied parent report of improvement in social awareness (P1), social cognition (P1), social communication (P1), autistic mannerisms (P1, P4) 9. Some worsening based on parent report; autistic mannerisms (P3), social cognition (P4) & social communication (P3, P4)	1
Spencer et al. (2008)	Time spent on playground			1

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Table 3 (*continued*)

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
		Teachers recorded time on & off playground with fellow classmates & second grade students.	1. Improvement in minutes spent on playground during intervention & maintenance	
Social groups (includes peers and siblings)				
Ashbaugh et al. (2017)	Social integration (community and extracurricular); Number of peers interacted with; Academic performance; Social validity	Number of social activities and peers interacted with were coded through a daily social (self-report & verified by peer mentor); Academic performance evaluated via GPA; Satisfaction of socialization rated through self-report	1. All participants increased in the number of social community and extracurricular activities per week; Maintenance was observed for P3 for community activities and extracurricular. P1 and P3 showed some maintenance in informal social activities 2. All participants increased in the number of peers interacted with. Data was cumulative and continued to demonstrate small increases post intervention 3. All subjects improved in their GPA 4. All participants reported increases in their social satisfaction with college and social activities	4
Baker (2000)	Play with siblings (% time in social play, Attention behaviors, child affect, % intervals engaged in ritualistic activity, & social validity)	Videotapes coded by naïve observers - 10 min probes in 30-40 min. play sessions Sibling interview Parent likert scales	1. All Increase in social play behavior (with maintenance) 2. All increase in positive joint attention behaviors (with maintenance) 3. All increase in affect ratings (with maintenance) for participants & siblings 4. Decrease in thematic ritualistic behaviors with maintenance for P1 & P2 (P3 did exhibit behaviors in 2 of 14 follow-up sessions) 6. Parent rated decreases in obsessional behavior for P1 & P3; P2 decreased then up slightly at follow-up; Parent rated how problematic activities were – P1 decreased, P2 stable, P3 decreased then slightly up at FU; Parent rated amount of sibling play – P1 increased, then decreased; P2 slight increase; P3 increase 7. All siblings increased in interest in playing with sibling	2
Baker et al. (1998)	Social behavior (% time engaged in social play in obsession & non-obsession theme & child affect)	Video tapes coded for percent of 10-sec intervals with appropriate social play & Affect rated with Affect Scale	1. P1 & P2 increased in social interaction with maintenance & FU; P3 maintained 100% at intervention, decreased slightly at maintenance & follow-up 2. P1, P2, & P3 increased in affect & maintained	1
Boyd et al. (2007)	Social behavior (% of intervals that child chose CI v. LP in choice % time child & peer engaged in social interactions amount of session time before child initiates to peer in AT Social Validity of appropriate play)	Data collected during classroom observations by graduate students Social validity rated by autism expert	1. Target child choices – P1 chose peer with CI about half the time; P2 chose peer with CI always; P3 chose peer with CI most of the time 2. Percent social interactions – higher during CI condition for all participants 3. Latency of initiation – P1 & P2 lower during CI; P3 never initiated during LP 4. Expert rated higher appropriate play with CI, lower inappropriate play with CI, & higher play frequency with CI for all participants	3

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Table 3 (*continued*)

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
Koegel Vernon et al. (2012)	Social engagement; Initiations	Percentage of intervals engaged with TD peers & initiations to TD peers coded from videotapes	1. Clear improvement for P1 & P2. Short intervention period for P3 so less clear 2. Increase in initiations for P1 & P2, but unstable for P1. Very minimal improvement in P3	3
Koegel and Fredeen et al. (2012)	Engagement with peers; Unprompted verbal initiations	Engagement with peers & unprompted verbal initiations to peers coded in vivo during each session	1. P2 & P3 showed immediate improvement in engagement with peers; P1 did not show stable improvement until intervention delivered in school rather than camp 2. P1 & P3 demonstrated increased in social interactions; P2 data less stable; P1 data more stable when in school rather than camp	2
Koegel and Ashbaugh et al. (2013)	% intervals engaged with peers; Frequency of initiations to peers; Social validity	Percentage of intervals engaged with TD peers & initiations to TD peers coded from videotapes; Social validation collected through 7 item self-report measure	Cohort 1: 1. P1, P2 & P3 showed increase in % intervals engaged with TD peers; Only P3 showed generalization. P4 showed minimal change 2. P1, P2 & P3 showed increase in initiations TD peers; P2 & P3 showed some generalization; P4 showed minimal change 3. Similar responses to TD peers to social validation questions Cohort 2: 1. All participants showed increase in % intervals engaged with TD peers; Generalization not observed 2. All participants showed increase in initiations to TD peers; Some generalization observed for P6 3. Similar responses to TD peers to social validation questions	3
Koegel and Ashbaugh et al. (2013)	Social activities; Quality of life; Satisfaction of socialization	Social activities were coded through a weekly activity log (self-report & verified by peer mentor); Quality of life evaluated as a social validity measure – type & quality of social interaction, academic achievement & employment activity; Satisfaction of socialization rated through self-report	1. All participants increased in the number of social activities attended; Evidence of maintenance following intervention 2. Qualitative reports of quality of life suggest vast improvement for all participants 3. All participants improved in their reports of satisfaction with college, peer interactions & friendships	2
MacCormack (2016)	Rate of initiations; Level of engagement; Affect	All variables coded as the frequency or score during 30 second interval coding.	1. Rates of initiations increased in structured and free play. Most improvement for P2. 2. Level of engagement increased for all participants. Effect strongest during structured play. 3. Slight improvement in affect, but less clear compared to other variables. Differences between structured and free play were small, but higher affect during free play.	1

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Table 3 (*continued*)

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
Activity/task based				
Adams (1998)	Primary outcome variable: Task score (# items completed in allotted time) Secondary outcome variables: Time off task, # of redirects, & Time spent engaging in self-stimulatory behavior	Videotapes coded by blind RAs	Data across all 6 tasks were variable & unstable Task score: 1. P1, P3 & P5 variable improvement across tasks but not stable. 2. P2 minimal/unstable change 3. P4 Improvement in some tasks; Worsening in Task C Off task & Redirects: 1. P1 & P3 spent more time off task & required more redirects unrelated to intervention 2. P2 & P5 less time spent off task. 3. P4 little change in off-task behavior (already low) Self-Stimulatory Behaviors: 1. P1 clear evidence of decrease in self-stimulatory behaviors 2. P2, P4 & P5 engaged in more self-stimulatory behavior during intervention: Data very unstable 3. P3 some evidence of decrease (Task C)	
Carnett et al. (2014)	Behavior (challenging & on-task)	Videos coded using partial interval coding for challenging behavior & whole interval coding for on-task behavior	1. Both token economies (with & without CI) increased on-task behavior, but on task behavior higher with CI 2. Both token economies decreased challenging behaviour, but lower challenging behavior with CI 3. Effects in inclusion classroom for generalization	3
Dunst et al. (2010)	Learning & learning opportunities	Parents rated number of different learning opportunities per week on a 5 point scale, frequency of child participation with activities on a 5 point scale & child learning/amount child benefitted on a 6 point scale	All participants/Group comparisons: 1. Parents in high interest group reported more learning activities 2.Parents in high interest group reported more frequent child participation 2. Parents in high interest group reported children benefited more from learning activities	0
Dunst et al. (2011)	Language, cognition, social, & motor developmental quotients	Developmental observational checklist system (DOCS) filled out by parents	All participants/Group comparisons: 1. High interest more progress than low interest group 2. 84% planned activities occurred 3. Difference in SES between groups (high interest → middle class; low interest → upper middle class) 4. All DQs increased in high interest except motor 5. Interest scores alone accounted for 28% variance in growth curve estimates	1
El Zein et al. (2016)	Reading comprehension	Observational coding of responses to reading comprehension questions & number of words uttered during oral retell	1. Reading comprehension responses to questions increased during CI condition 2. Increase in number of words uttered increased during CI condition 3. Both measures more stable during CI condition	2
Koegel et al. (2017)				

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Table 3 (*continued*)

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
	Socialization (Activity engagement; Social engagement; Affect/interest in activity; Initiations to peers); Social validation	Activity engagement, social engagement, affect/interest and initiations all coded via videotapes using partial interval coding. Social validation rated through surveys with participants and peers.	1. Both P1 & P2 showed immediate improvement in engagement within activities with a positive history and engagement remained high (P1: 96.9%; P2: 85%). Both P1 & P2 showed no improvement in engagement for negative history. 2. P1 showed immediate, but moderate, improvement in social engagement with peers within activities with a positive history. Engagement was varied for P1 over the course of the sessions. P2 showed immediate improvement, but this decreased sharply as the study progressed. Neither P1 or P2 showed improved social engagement for negative history activities. 3. Both P1 & P2 showed improvement in initiations to peers in positive history activities, however this was varied and decreased over the course of the study. No improvement in initiations for negative history activities. 4. Child affect/interest improved and remained high for P1 during positive history activities. P2 demonstrated some immediate improvement in affect/interest for positive history activities, however this was not maintained. Affect remained negative for both subjects for negative history activities. 5. Both subjects reported positive experiences with the positive history activities.	
Kryzak and Jones (2015)	IJA; CI intensity; Generalization of IJA across partners, settings & non-CI activities	IJA coded from video recorded sessions; Parent rating scale of CI intensity	1. Percentage of IJA increased for all participants but data unstable 2. Reduction in redirecting/interrupting CI in P2 & 3. No 3change for P1. Decrease in CI related social-interference in all participants; P3 increased time spent in CI-related activities 3. Minimal evidence of generalization to partner or mastery in P2 & P3	3
Kryzak et al. (2013)	RJA; CI Intensity; Socialization; Generalization	RJA coded from video recorded sessions; Parent report of CI intensity & social interaction	1. Increased in percentage of RJA increase but data unstable for P2 & P3 2. CI intensity reduction in P2, CI intensity stable or increased slightly in P1 & 3 3. Most ratings of social interaction show improvement 4. Some evidence of generalization	3
Marshall (2017)	Reading comprehension	Reading comprehension variables (oral retell, word count per minute [WCPM], & reading comprehension questions) coded from audio recordings of sessions.	1. No differences in WCPM for P1 or P2 across different conditions. Data very variable. 2. Slight trend for increasing relevant word in P1, but not specific to condition. No differences for P2. 3. Slight increase in percentage of correct questions for P1	3

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Table 3 (*continued*)

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
			in frequent condition, but very variable. No differences for P2.	
Trivette and Dunst (2011)	Positive & negative child affect; Child social responsiveness	Affect measured via investigator-administered scale that rated 5 indicators of positive affect on a 5 point scale; Child social responsiveness (7 social behaviors) was rated by caregivers on a 5 point scale.	All participants: 1. Medium to large effect sizes for negative child affect & social responsiveness 2. Small effect size for positive affect 3. Children demonstrated more positive & less negative social-affective behaviors Group comparisons: 1. High interest-based group showed more positive affect, less positive affect & more social responsiveness than low interest-based group	0
Motivational PRT				
Vismara (2005)	Type/quality of IJA gestures (gaze alternation, pointing, giving, showing, vocalization, combination)	Gestures coded from videos using Noldus Observer.	1. P1 & P3 showed increases in several types of IJA gestures; P2 did not show immediately; All children improved in use of multiple gestures	5
Vismara and Lyons (2007)	IJA (number of IJA initiations, contingencies to IJA); Parent-child interactions	IJA behaviors & parent-child interactions coded from videos of sessions.	1. IJA increases clear & significant for P1; Some effect for P2 in 1st half of study; P3 less clear – more IJA with non-preferred stimuli 2. Parent-child affect data more stable for CI conditions than non-CI for all children; All improved from baseline	4
Video modelling				
Haymes (1995)	Conversation skills (Verbal initiations; Verbal Responses; Verbal Elaborations; Appropriate peer-directed speech; Percentage of conversation; MLU; Nonverbal initiations; Cooperative play) Social validity	Video tapes transcribed & coded for conversation skill variables. Social validity by parents & peers	1. All participants increased verbal initiations with peers following video model & generalized; Effects did not differ based on condition (CI vs. non-CI) 2. P1 & P3 increased verbal responses that were generalized; P2 variable 3. P1 increased & generalized verbal elaboration; Slight increase for P2; P3 variable 4. P1 & 3 increased peer directed phrased & generalized; P2 increased slightly 5. All participants showed increased in percentage of conversation; Change not a great for P1 6. P1 & 2 MLU increased slightly & generalized; P3 increased but no generalization	0
Jung and Sainato (2015)	Engagement with game & Engagement with peers Inappropriate Behavior Social validity	Coded video tapes. Social validity rated by teacher	1. P1, P2, & P3 increased verbal & nonverbal engagement with games after VM & at FU & generalized 2. P1, P2, & P3 increased verbal & nonverbal engagement with peers after VM, but P3 less substantially 3. P1, P2, & P3 decreased in inappropriate behavior after intervention & maintained 4. Social Validity – teachers rated meaning & feasible/ acceptable	3

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Table 3 (*continued*)

Study	Outcome Variable	Outcome Measure	Results	Risk of Bias (# of low ratings)
Ohtake et al. (2015)	Bathroom skills: Drying hands; Arranging shoes; Covering buttocks; Tucking shirt	Behaviors coded from videotaped sessions. Scores based on 0 to 3 rating scale	1. Immediate improvement in drying hands for at least 5 seconds & maintenance observed in 9 out of 12 sessions 2. Improvement in arranging shoes to level 2/3 after VHM introduced; Generalization observed in 10 out of 12 sessions 3. Improvement in covering buttocks took longer to reach level 3 but maintenance observed in 6 out of 6 sessions 4. Immediate improvement in tucking shirt & maintenance observed in 4 out of 4 sessions	4
Social conversations				
Lepper et al. (2017)	Percentage of intervals with conversation; conversation topic	Conversations coded during 10 second intervals	1. Higher percentage of conversations during contingent (experimenter attention) condition 2. Both participants increased in CI-unrelated conversation. Both participants decreased in CI-related conversation	4

CI = circumscribed interest; FU = follow up; GPA = grade point average; IJA = initiation of joint attention; LP = low preference; MLU = mean length of utterance; P = participant ; RJA = response to joint attention; VHM = video hero modelling; VM = video modelling.

3. Results

3.1. Study selection

We located 23,236 records following a thorough search of databases as well as records identified through other sources such as hand searching (Fig. 1). After duplicates were removed, 14,578 records remained that were then screened based on their title and abstract. 246 full-text articles were assessed for eligibility and 215 were excluded because they did not meet eligibility criteria, leaving 31 studies for data extraction.

3.2. Study design

Of the 31 articles, the majority were single subject designs (k = 28; Table 1). There was one group design (Quasi Experimental), with data from the same sample published in three articles (Dunst, Trivette, & Masiello, 2011; Dunst, Trivette, & Masiello, 2010; Trivette & Dunst, 2011). The majority of studies were published after 2009. Six studies were dissertations.

3.3. Age of subjects

Six studies included children that fell into multiple age categories. The majority of studies were focused on toddlers/preschool (k = 13/31) or school-aged children (k = 17/31). Six studies included adolescents and two focused on adults (Table 1). Eighteen

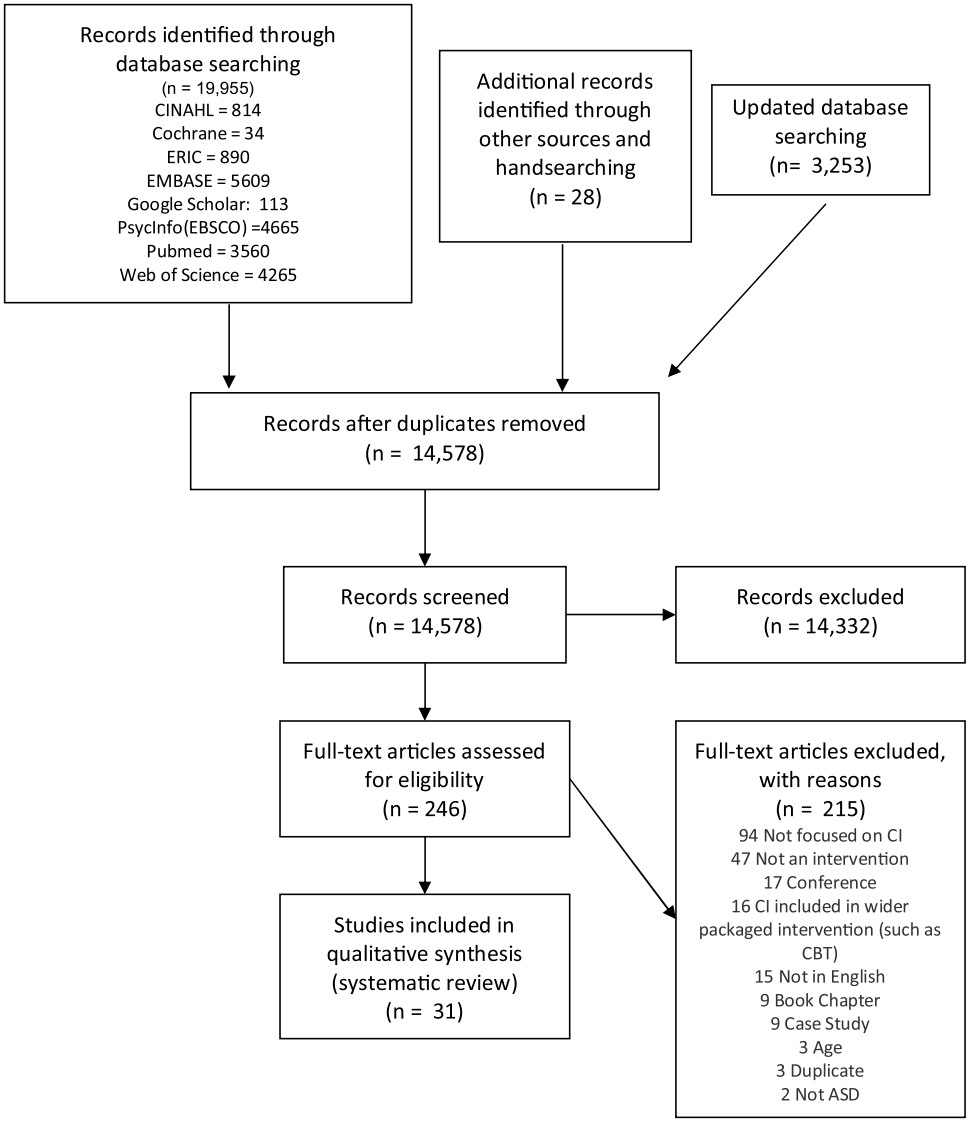

Fig. 1. Flow diagram of studies included in systematic review.

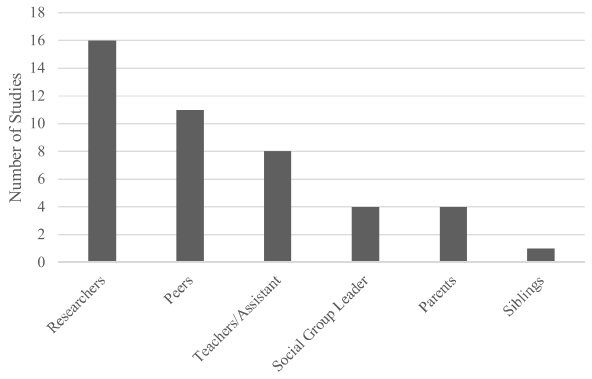

Fig. 2. Delivery Method.

females were included as participants across the 31 studies (28 studies reported in 31 published articles/dissertations) and a total of 92 *unique* participants (the 17 in the Dunst et al studies were only counted once as were the 3 in Vismara (2005) and Vismara and Lyons (2007).

3.4. Types of interests

The most common type of CI utilized in treatment was TV Shows or Movies (N = 21), followed by Popular Characters (N = 18), computers/video games (N = 12) and transportation (N = 12; Table 1). Interests were typically confirmed by parent or teacher interviews/report (N = 22). Eleven studies confirmed interests through child observations and five with a preference assessment (Table 1). Six studies asked the participant what their interest was. One study reported that the intervention was individualized to the participant's interests and confirmed interests through self and parent report, but did not state what the individual interests were in the text of the article (Ashbaugh, Koegel, & Koegel, 2017).

3.5. Intervention approach and delivery method

The 31 studies ranged in intervention approach (Table 2). Six broad categories were identified. Nine of the 31 interventions were implemented during social groups (including peers and siblings). Six (out of 31) studies were implemented with power cards. Other approaches used included video modelling (k = 3) and activity/task based interventions (e.g. CI were embedded into already occurring routines and activities such as reading activities at school, and daily activities at home ; k = 10). One study used embedded CI within conversation training and bids (Lepper, Devine, & Petursdottir, 2017) and two studies used Pivotal Response Training (PRT) – a naturalistic intervention based on ABA principles and developmental approaches that focus on improving motivation during learning and socialization opportunities (Vismara & Lyons, 2007; Vismara, 2005).

The majority of interventions were delivered by trained researchers (k = 14/31; Table 2; Fig. 2). This was followed by teacher/teaching assistant implementation (k = 7/31) and peers (k = 9/31). Ten studies had more than one means of intervention delivery (e.g., parents or assistants and peers/peer mentors). Four studies included technology (videos or video games) as a mode of implementation. Power cards were most likely to be implemented by researchers or teachers/classroom staff. Social groups, particularly for adolescents, were often implemented by peers, whereas play-based interventions and those targeting social communication (such as joint attention) were often delivered by parents.

3.6. Outcome variables, results and risk of Bias

To aid interpretation, Table 3 is organized by the Intervention approach (*how* the intervention was delivered) as defined by the six categories previously mentioned. The majority of studies reported outcomes pertaining to social/communication skills (k = 22/31; Table 3). These behaviors ranged from play with peers, conversation skills, social activities and joint attention (Table 3). Five studies reported *on task* behaviors, such as direction following and completing assigned tasks. Remaining outcome variables included sportsmanship (n = 1), resiliency (k = 1), reading comprehension (k = 2) and adaptive behavior (toileting; k = 1). Five studies reported the social validity and acceptance of the intervention from parents and/or teachers. One study reported the social validity of the intervention from the participants and their peers (Koegel, Oliver, & Koegel, 2017).

For social group interventions, 7 out of 9 studies reported mostly positive findings across subjects and/or variables (Fig. 3). Of the studies that reported mostly positive outcomes (k = 7), the risk of bias was mostly rated as low or unclear for most categories, with the exception of MacCormack (2016) that was rated as high risk of bias in four out of seven categories Fig. 6 – Supplementary). The remaining two social group interventions reported somewhat mixed findings, with one participant showing slower (Koegel, Kim, Koegel, & Schwartzman, 2013) or little improvement (Koegel, Vernon, Koegel, Koegel, & Paullin, 2012) resulting from the intervention. These studies were rated as unclear risk of bias (Figure 6 – Supplementary). Five studies (out of 9) reported maintenance and/or generalization. Of these five, four were rated as positive (Ashbaugh et al., 2017; Baker et al., 1998; Baker, 2000; Jung &

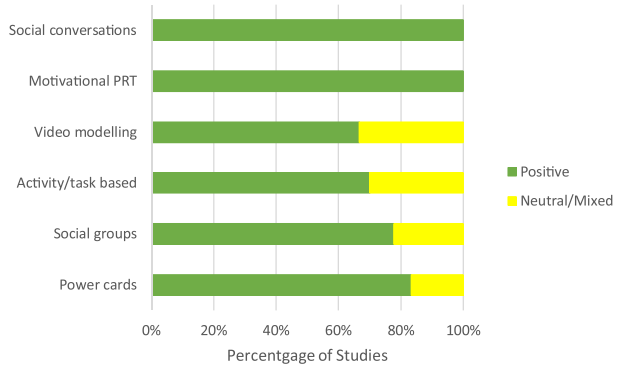

Fig. 3. Study Outcome (Positive or Neutral) by Intervention Approach.

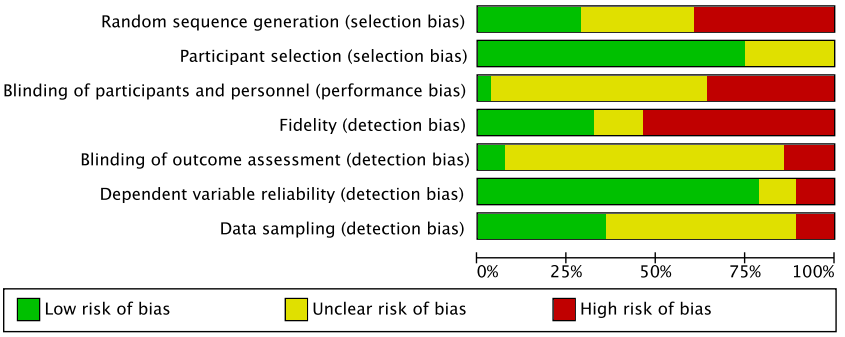

Fig. 4. Risk of Bias: Single Case Designs.

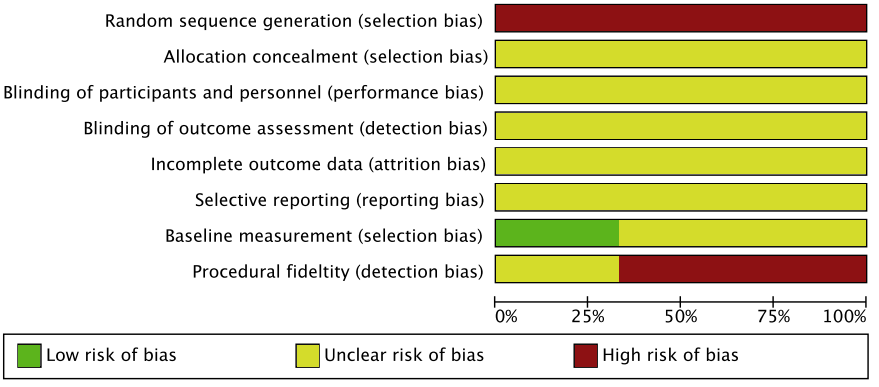

Fig. 5. Risk of Bias: Group Design.

Sainato, 2015) and one as negative (i.e. no maintenance and/or generalization; Koegel, Ashbaugh, Koegel, Detar, & Regester, 2013).

For the power card interventions, 5/6 studies reported mostly positive results across participants and/or variables. Of these 5 studies, 2 received mostly low ratings for risk of bias (Angell, Nicholson, Watts, & Blum, 2011; Campbell & Tincani, 2011) and 2 received primarily high ratings (Keeling, Smith Myles, Gagnon, & Simpson, 2003; Spencer, Simpson, Day, & Buster, 2008; Fig. 6 – Supplementary). One power card intervention study reported mixed findings (Kuligowski, 2010). Of the four power card studies that reported maintenance and/or generalization, one reported positive change over time (Spencer et al., 2008) and three were rated as mixed (Campbell & Tincani, 2011; Davis, Boon, Cihak, & Fore Iii, 2010; Keeling et al., 2003).

Of the ten activity/task-based intervention, seven reported positive results (2011, Carnett et al., 2014; Dunst et al., 2010; El Zein, Solis, Lang, & Kim, 2016; Koegel et al., 2017; Kryzak & Jones, 2015; Trivette & Dunst, 2011) and three mixed results (Adams, 1998; Kryzak, Bauer, Jones, & Sturmey, 2013; Marshall, 2017). Of the six studies that reported positive outcomes, most had mostly mixed/unclear ratings of bias (Fig. 6 – Supplementary). One study reported positive effects on maintenance and/or generalization (Carnett et al., 2014) and another reported no generalization effects (Kryzak & Jones, 2015).

Of the three video modelling studies, two reported mostly positive outcomes (Jung & Sainato, 2015; Ohtake, Takahashi, & Watanabe, 2015) and one reported mixed findings (Haymes, 1995). Ohtake et al. (2015) received mostly low risk of bias ratings,

whereas Haymes (1995) and Jung and Sainato (2015) were more mixed/unclear.

The one intervention delivered via social conversations (Lepper et al., 2017), reported mostly positive outcomes and received mostly low risk of bias ratings (Fig. 6 – Supplementary). The two motivational PRT studies reported positive outcomes (Fig. 3) and mostly low risk of bias ratings (Fig. 6 – Supplementary).

3.6.1. Quality assessment and risk of bias

71% of the SSD studies were rated as either *unclear or high risk of bias* for random sequence generation, suggesting a potential selection bias effect across studies (Fig. 4). The majority of studies were rated as either *unclear or high risk of bias* for both blinding of participants and personnel and blinding of outcome assessment. However, the majority of studies were rated as *low risk of bias* for both participant selection and dependent variable reliability (Fig. 4). Just over a third of studies were rated as *low risk of bias* for fidelity and data sampling.

The three published studies stemming from the same sample (Dunst et al., 2010, 2011, Trivette & Dunst, 2011) were rated as *high risk of bias* for random sequence generation (Fig. 5). Two studies were rated as *high risk of bias* for procedural fidelity and one as *unclear risk of bias*. The majority of variables were rated as *unclear risk of bias* for all three studies (Fig. 5). Only one study was rated as *low risk of bias* for baseline measurement.

4. Discussion

Overall, the findings from this systematic review indicate that harnessing CI within interventions have potential for positive change for individuals with ASD. Across a number of predominately SSD studies, improved outcomes were observed in a variety of domains, including adaptive behavior (Ohtake et al., 2015), social-communication (Baker et al., 1998; Vismara & Lyons, 2007) and play (Baker, 2000). Of the studies that reported social validity, parents and/or teachers reported high levels of satisfaction and interventions were rated as feasible, suggesting that interventions incorporating CI would be welcomed by caregivers within the ASD community. Further studies are needed to definitively determine whether it is the CI or other aspects of the intervention package that are active ingredients for promoting positive change.

Neither intervention approach (*how* the intervention of implemented) nor delivery method (*who* implemented the intervention) appeared to impact child outcomes in a consistent way. It may appear that PRT or social conversation approaches led to more positive outcomes (Fig. 3) but the number of studies using these approaches were smaller compared to other more common approaches, such as social groups and activity-based interventions. Who delivered the intervention also did not seem to influence outcomes. However, it is noteworthy that parents (N = 4) and siblings (N = 1) were underutilized in the interventions, suggesting the importance of further research to examine the important role of families in the delivery of interventions that embed CI.

While most studies reported improved outcomes, significantly fewer studies reported maintenance or generalization of findings. When maintenance and/or generalization data were reported, results were less consistent. This suggests that the effects of CI-based interventions may decrease in their potency over time and/or may not naturally transfer to other people or settings. It is possible that booster interventions are needed to maintain effects or that multiple caregivers or providers should be trained to support generalization. Of course, it is also possible that intervention effects diminish over time because the children's CI may change, as they could simply become interested in other things.

The majority of reviewed studies used single subject designs. This was anticipated given the highly personalized nature of these intervention approaches. Given the small sample sizes that are inherent to SSD and the lack of a separate control condition, it is more difficult to draw generalizable conclusions from these studies. Only one group design study (reported across three published papers) was included in our review. The series of Dunst and colleague articles interviewed parents to identify the child's interests and opportunities to embed learning opportunities around these interests; however, the individual interests of each child were not reported likely because of the larger sample involved in a group design study. While the majority of manualized interventions can be adapted to meet individual needs (e.g. Dawson et al., 2010; Kasari, Gulsrud, Paparella, Hellemann, & Berry, 2015; Mesibov, Shea, & Schopler, 2005; Smith, 2001), it is unclear, and often not explicitly stated, how these interventions incorporate individualized interests, if at all. Given the drive toward personalized medicine approaches in ASD, the role of CI is likely to represent an important one as we begin to target interventions to individual behavioral (or genetic) profiles. Further, incorporating interests within intervention and designing interventions around these interests represent one of the only strength-based approaches currently used within ASD behavioral treatments (Bellini & McConnell, 2010; Mottron, 2011); therefore, including more individualized CI as components within large-scale and/or manualized treatments should be prioritized.

Regarding participant characteristics, 19% of participants were female, corresponding with the male: female diagnosis ratio (4:1). Recent research suggests that females with ASD hold different CI and may engage with these in different ways (Hiller, Young, & Weber, 2014; Sutherland, Hodge, Bruck, Costley, & Klieve, 2017). Thus, it is important to consider how using CI within interventions may lead to different intervention approaches and outcomes for males and females, as we cannot simply assume that females are motivated by the same interests as males (such as transportation and sports).

Despite the opportunity, only a handful of studies (k = 7) examined the benefits of CI-based social groups and activities for adolescents and adults. Reichow and Volkmar (2010) also found a significant lack of research examining social skills interventions in general for individuals with ASD in this age range. Given this population appears to remain understudied, coupled with the increasing numbers of individuals with autism in this age range, this could be an area of great promise, especially examining ways in which to promote improved quality of life as individuals enter adulthood and beyond. As CI often represent *islands of ability* for individuals with ASD, this strength-based approach could directly lead to social and employment opportunities. Adults with a diagnosis of ASD

are less likely to be engaged in employment, post-secondary education and leisure activities (Taylor & Seltzer, 2011). Therefore, utilizing and capitalizing upon CI represents one approach to increase the low rates of employment and community engagement, while also providing adults with a sense of belonging. The potential of using CI to gain meaningful employment has been discussed in the popular media and the field (Attwood, 2003; Hendricks, 2010; Klin et al., 2007), but to our knowledge has not been systematically studied.

The majority of studies confirmed individuals' CI with teachers or family members and, in some cases, the participants themselves. A number of studies combined multiple confirmation approaches, such as including preference assessments. Multiple approaches are seen as beneficial, especially for minimally verbal children, who may not be able to directly report their CI. However, there is still a need to develop a common, systematic approach across studies for confirming CI in order to ensure comparability. Perhaps the inclusion of physiological measures, such as eye tracking (Sasson & Touchstone, 2014; Sasson et al., 2008), could be added as an objective measurement of CI given the current reliance on informant or self-report.

Finally, a handful of studies measured how the intervention impacted the child's engagement with their specific interests, exploring potential negative implications of CI-based interventions. Of the studies that reported interference of, engagement with, or expertise in the CI, the findings were inconclusive (Kryzak & Jones, 2015; Kryzak et al., 2013; Lepper et al., 2017). Future research would benefit from the inclusion of measures designed to not only identify CI, but also measure the impact (positive and negative) of interests. Examples include the Interests Scale (Bodfish, 2004; Turner-Brown et al., 2011) or the Survey of Favorite Interests and Activities (Smerbeck, 2017), which could be easily implemented pre and post intervention to assess change and provide a more standardized way of measuring engagement with CI and also their impact on daily life.

4.1. Future directions: implications for research and practice

As incorporating CI within treatment represents a highly individualized approach to intervention, this inevitably leads to a small number of group designs to examine intervention effectiveness. Yet, to fully understand the impact of this individualized approach to treatment, more group designs are required to be able to conduct an actual meta-analysis and understand the magnitude of treatment effects. While it is common to aggregate effects of studies employing similar methods of implementation or utilizing similar outcome measures, due to the wide age range of this review, the range of implementation methods, and outcome variables, this was not a suitable method. For example, there were two reading comprehension studies with a total of three participants aged 8–17 years. Aggregating effects across these two studies would not have provided a meaningful statistic above pure description of the studies. Replication of SSDs will better allow for the aggregation of results to produce meaningful effect sizes across studies.

Another caveat of the high number of SSD is the high risk of bias and high variability across studies. For example, El Zein et al. (2016) and Marshall (2017) reported similar reading comprehension studies across different age ranges with very different outcomes. Therefore, replication of studies is required to understand the appropriateness of treatment designs and the effects of interests as active ingredients. While SSDs allow us to examine in detail the individual effects of an intervention and are often extremely high-quality studies, they can be accompanied by a high risk of bias, often because of the lack of randomization and small sample sizes. The association between risk of bias and positive outcomes was not clear cut: positive outcomes did not appear more likely from poorer quality studies, particularly for SSD, and a number of studies rated with unclear risk of bias produced mixed results.

It should be noted that a handful of studies did not state the individual participant's interests. While this is a flaw of these individual studies, the decision was made to include these as they clearly specified in the study design that interventions were designed to include individualized, CI and these were confirmed by the study team or parents/teachers. As discussed previously, more standardized and rigorous methods of confirming CI, both within interventions and wider research, should be implemented to allow for comparison of different intervention approaches.

More group designs are required to understand the potential impact of CI within intervention. Group designs typically often have greater generality, external validity and less selection bias, which would reduce such risks, and would provide more breadth of knowledge for how these interventions are likely to work for a larger number of individuals with ASD. Only one group design (reported across three published articles) was included in this review. These studies received mostly unclear ratings for risk of bias and high risk of bias for random assignment; therefore, conclusions about whether CI are valid *active ingredients* within this specific intervention are not possible. Instead, future research should include randomized CI vs. non- CI conditions to understand whether it is the inclusion of these interests, rather than other factors such as mode of delivery, that serves as a vehicle for change.

In addition, more information is needed about how static or changing CI are and whether interventions need to vary based on the type of interest held. For example, an intense interest in cars may be successfully embedded within a play-based intervention, but could be harder to embed within a social group model. Given both the positive and negative implications of CI (Mottron, 2011; Turner-Brown et al., 2011), it is also unclear whether researchers should encourage these interests, help develop new ones or fade these once skills have been acquired. As discussed previously, despite the benefits, there were only a handful of studies utilizing CI as a tool in higher education and employment. This lack of published studies is striking and an area where future research should focus, particularly with the current focus on transition age individuals with ASD (van Schalkwyk & Volkmar, 2017; Volkmar, Jackson, & Hart, 2017).

As noted in the introduction, a number of researchers have used physiological and neurophysiological methods to examine both positive and negative implications of CI on cognitive processes, such as attention, motivation and task performance (Benning et al., 2016; Cascio et al., 2014; Foss-Feig et al., 2016; Sabatino et al., 2013; Sasson et al., 2008; Unruh et al., 2016). As with a number of early-intervention and behavioral studies (Dawson et al., 2012; Yang et al., 2016), combining traditional forms of outcome measurement, such as parent report and observation, with more novel tools would enable us to understand potential effects of treatments

incorporating interests at a neural level, and whether these treatments lead to enhanced expertise or have an adverse effect on the allocation of attention.

4.2. Limitations

There are a handful of limitations that are worthy of discussion. We did not include any articles or dissertations that were not published in English. While it is unlikely this had a large impact on the studies and data extracted in our final review, it is possible a handful of eligible studies may have been excluded. While most studies reported confirming individual interests through parent report, preference assessments or subject interviews, a handful of studies did not report how CI were confirmed or report what the individualized CI were, therefore, creating some uncertainty as to whether it was the participant's actual interest or simply a highly preferred item or object and reducing the replicability of these studies. This was especially true for the collection of Dunst and colleague articles (2010, 2011), Trivette and Dunst (2011). Thus, a trade off with larger, group studies is the lack of individual details that SSD can provide.

Further, we did not assess individual study quality for SSD prior to inclusion and data extraction, using methods such as the What Works Clearinghouse Criteria (IES, 2012); instead, we included all studies and reported risk of bias. While some included studies could have been poorly designed, the majority reported positive outcomes, and therefore are likely to have met most basic quality indicator guidelines.

Finally, we did not contact individual authors of included or excluded studies in order to include grey or unpublished literature, which may slightly bias our findings. While we did include unpublished dissertations, this only accounted for 19% of our included studies. Although inclusion of dissertations should limit the threat of publication bias, reliance on this method alone to account for all publication bias is not adequate and we cannot rule out the possibility of publication bias in the current review.

5. Conclusions

Circumscribed interests in ASD often represent *islands of ability* for individuals. To date, very few studies have modified or developed interventions to include these strengths. Results from the 31 studies included in this systematic review suggest that the inclusion of CI within interventions can lead to positive effects in a number of domains, including adaptive functioning, communication and interaction with peers. More research is required to understand how interests can best be incorporated within large-scale trials in order to avoid an over-reliance on SSD. Future research ideas include examining the effects of these personalized interventions using methods such as eye tracking and EEG and further utilization of CI in employment and education services for adolescents and adults on the spectrum.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.rasd.2018.09.008.

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