Cognitive training as a resolution for early executive function difficulties in children with intellectual disabilities

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Abstract

Core executive functions (EF) such as attention, and working memory have been strongly associated with academic achievement, language development and behavioural stability. In the case of children who are vulnerable to cognitive and learning problems because of an underlying intellectual disability, EF difficulties will likely exacerbate an already compromised cognitive system. The current review examines cognitive training programs that aim to improve EF, specifically focusing on the potential of this type of intervention for children who have intellectual disabilities. We conclude that despite considerable discrepancies regarding reported intervention effects, these inconsistencies can be attributed to flaws in both program and study design. We discuss the steps needed to address these limitations and to facilitate the advancement of non-pharmaceutical interventions for children with intellectual disabilities.

Key Words: Executive Function; Attention; Working Memory; Intellectual disability; Cognitive Training; Children

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Concentrating on a task, switching attention between tasks, or inhibiting impulsive responding are critical components in the development of cognitive control. Collectively known as executive functions (EF) these skills emerge early in life and their development becomes progressively more robust from the preschool years onwards (Steele, Karmiloff-Smith, Cornish, & Scerif, 2012; Zhan et al., 2011). The importance of EFs are highlighted by recent studies demonstrating that these skills are more strongly associated with school readiness than IQ (Blair & Peters Razza, 2007), serve as predictors of literacy and numeracy scores in preschool through high school (Clark, Pritchard, & Woodward, 2010; Clark, Sheffield, Wiebe, & Espy, 2013), facilitate social inclusion and peer relationships (Gomes & Livesey, 2008), and play an important role in maintaining mental health across the lifespan (Diamond, 2012; Meyer et al., 2004). Disruption to these essential processes can lead to increased levels of distractibility, impulsivity, forgetfulness and poor focus. In the case of children who are especially vulnerable to learning impairments, because of an underlying intellectual disability (e.g. Autism, Down syndrome), EF difficulties will likely exacerbate an already compromised cognitive system. The developing child’s ability to interact with the world around them is significantly impacted, reducing their capacity to engage in educational programs and increasing their already heightened risk of long-term behavioral and emotional problems (Einfeld et al., 2006; Hofer et al., 2009).

Treatment options for improving EF in individuals with intellectual disabilities (ID) are sparse and largely target only behavioural features such as inattention and hyperactivity. As a result psychostimulant medication comprises the most common treatment option, and although short term effects of drugs such as methylphenidate and amphetamine are well documented (e.g. Swanson, Baler & Volkow, 2011), less is known about the long term impacts of prolonged use. Findings also indicate that children with ID are less likely to show therapeutic benefits and have increased vulnerability to the negative side effects of psychostimulants than typically developing (TD) children (Aman, Farmer, Holloway & Arnold, 2008). In light of the shortfalls of current interventions, recent investigations have suggested that cognitive training may provide an adjunct to pharmaceutical interventions and a riposte to the concept that cognitive impairments are permanent. An increasing number of studies have explored the impact of EF training in TD children, including those within the general intellectual range of the TD population, such as children with Attention Deficit Hyperactivity Disorder (ADHD), and have reported promising effects on early working memory (Alloway, Bibile, & Lau, 2013; Thorell, Lindqvist, Bergman Nutley, Bohlin, & Klingberg, 2009) and attentional control skills (Rueda, Rothbart, McCandliss, Saccomanno, & Posner, 2005b). Given the high prevalence of cognitive difficulties in children
with ID, coupled with the significant lack of suitable interventions, an extensive investigation of how cognitive training can strengthen core EF skills in individuals with limitations in intellectual functioning is warranted.

In this narrative review, we examine current cognitive training programs, specifically those that target core EF, such as attention and working memory. Our primary aim is to assess existing cognitive training studies in the context of children with ID in order to gauge the potential benefits this intervention may offer in terms of cognitive and behavioural improvements for those with limited cognitive abilities. Although several narrative and meta-analytic reviews have assessed the effect of cognitive training in TD children and children with ADHD (Shipstead, Hicks & Engle, 2012; Shipstead, Redick & Engle, 2012; Klingberg, 2010; Morrison & Chein, 2011; Melby-Lervag & Hulme, 2013), to our knowledge this is the first review to examine the potential application of cognitive training in children with ID.

1. Executive Functions: Target Training Domains

1.1 Attention

The attention system is complex and multifaceted, and as such interacts with several cognitive domains (Posner & Petersen, 1990). Despite previous conflict regarding the definition of attention there is now widespread agreement that there are three separable cognitive components (Fan, McCandliss, Fossella, Flombaum, & Posner, 2005; Posner & Petersen, 1990) that are in place by as young as 6 years of age (Manly, Anderson, Nimmo-Smith, Turner, Watson & Robertson, 2001). These processes are selective attention, i.e. the ability to selectively attend to aspects of the environment; this network is operated by parietal structures connected with frontal eye fields and the superior colliculus. Sustained attention i.e. the ability to sustain attention on a task over time and maintain a high state of sensitivity to incoming information; this involves the parietal cortex, right frontal cortex and locus coeruleus. And lastly executive attention, the ability to control attention on a fixed goal while ignoring conflicting information; this process involves the left and right frontal areas as well as the anterior cingulate cortex.

All three attentional processes operate through a series of distinct yet complementary neural circuits, and undergo dramatic postnatal development in line with the maturation of each of the associated neural structures (Ruff & Rothbart, 1996). Due to the significant overlap between the attentional networks, disruption to one network is likely to have a significant and detrimental knock-on effect to the subsequent networks. For instance weaknesses in inhibitory control, a process associated with executive attention which allows us to regulate our behavior and attention would likely result in subsequent weaknesses in tasks requiring selective
attention, due to an inability to suppress attention to irrelevant or distracting stimuli. Therefore, successful mastery of each attentional network is imperative for subsequent cognitive development (Diamond, 2013), as well as behavioural control (Moffitt et al., 2011) and academic achievement (Fisher, Thiessen, Godwin, Kloos, & Dickerson, 2013). These insights highlight the potential of cognitive training programs that target attention in children by indicating that widespread improvements may occur in these associated domains. Furthermore, attention incorporates both behavioral and cognitive features and it is therefore important to consider this relationship when assessing interventions aimed at improving this system. Unlike pharmaceutical interventions that largely target behavioural features of attention (e.g. hyperactivity), cognitive training offers the potential to target underlying cognitive attention, thus facilitating improvements at the cognitive and behavioural level.

1.2 Working memory

Another core EF is working memory (WM) which is defined as the ability to maintain and manipulate information over short periods of time (Baddeley & Hitch, 1994). It is a key determinant of several higher-order cognitive functions such as reasoning, fluid intelligence, problem solving and language comprehension (Borella, Carretti, Riboldi, & De Beni, 2010; Nettelbeck & Burns, 2010). WM differs from short-term memory (STM) as it requires the manipulation of information, in addition to storage (Diamond, 2013). As such WM is shown to develop later and slower than STM (Davidson, Amso, Cruess Anderson, & Diamond, 2006). Despite the complex nature of WM, it is not an isolated module and has been closely related to attention, with the two domains increasingly being viewed as overlapping constructs (Cowan, Morey, AuBuchon, Zwilling, & Gilchrist, 2010; Gazzaley & Nobre, 2012). Indeed, both of these domains share similar neural regions within the prefrontal parietal system (Gazzaley & Nobre, 2012; Ikkai & Curtis, 2011; Nobre & Stokes, 2011). Furthermore, it has been suggested that the development of working memory capacity is reliant on the pre-established development of core attention skills (Kane, Bleckley, Conway, & Engle, 2001; McNab et al., 2008). It is probable that primitive attention networks that allow children to filter information, assist in ensuring that WM capacity is not overstrained (Zanto & Gazzaley, 2009).

Both WM and attention have been given research priority in the field of cognitive neuroscience due to their central role in cognition, learning and behavior. It is therefore unsurprising that these are the main functions targeted by current cognitive training programs.
2. Cognitive Training: The Concept

Cognitive training is based on the concept that repeated practice within a specific domain will result in gains in both cognitive and behavioural efficiency of the targeted domain, as well as subsequently transferring improvements to untrained domains. The premise for cognitive training can be further defined via comparison with physical training. For instance, engaging in cardiovascular exercise such as running, will produce improvements in an individual’s ability to run, however improvements are also likely to occur across other exercises, such as swimming or basketball that equally rely on the cardiovascular system.

Although traditionally cognitive constructs were viewed as stable, growing research demonstrates that cognitive and neural structures are highly responsive to environmental influence; that is that the developing brain has a large degree of plasticity (Teicher et al., 2003). Although plasticity has been shown in the adult brain (Maguire, Woollett, & Spiers, 2006) it is likely that this occurs via different mechanisms than in early childhood. Increased functional connectivity and specialisation of cortical circuits are emerging features of the developing cortex and it is therefore unsurprising that cognitive training has been shown to be most effective in early childhood (Wass, Scerif, & Johnson, 2012). In addition recent studies have refuted previous claims that individuals with ID experience cognitive stagnation and have instead highlighted that those with ID experience cognitive delay (Cornish, Cole, Longhi, Karmiloff-Smith, & Scerif, 2013). These findings indicate that cognitive weaknesses observed across ID’s are not necessarily permanent and that individuals with cognitive impairments may also benefit from interventions such as cognitive training.

3. Method

3.1 Search Strategy

To identify relevant research studies, we searched electronic databases (PsychInfo, Pubmed, Web of Science and the Cochrane Central Register of Controlled Trials) using combinations of the following search terms: children, cognitive training, cognitive intervention, attention training, working memory training. All searches were carried out in March 2014 and searches were limited to English-language and full length peer-reviewed articles. No other restrictions (e.g. publication date) were applied. In addition the reference lists of all included articles were hand-searched to identify any additional studies.

3.2 Inclusion Criteria

All study designs except single case reports were included in the review. Studies had to provide empirical data on performance before and after cognitive training, therefore articles that did not report their own
data such as review articles and meta-analyses were excluded. Importantly studies were only included if they described a training program that targeted cognitive domains, such as WM, attention, or broader EF, e.g. planning or inhibition. As such behavioural and social skills training programs were excluded. Although we wanted to be as comprehensive as possible in the literature search, our primary focus was to assess the potential of cognitive training in children (younger than 16 years), therefore adult studies were excluded. Studies also had to provide cognitive training to individuals who were either typically developing (TD), had a diagnosis or ADHD or an intellectual disability (ID). In order to be as inclusive as possible we included individuals with ID of different etiology (e.g. autism, fragile x syndrome, Down syndrome, and Williams syndrome).

3.3 Data Extraction

Data was extracted from eligible studies regarding the study design; sample size, sample population, training program, training procedure, control group, outcome measures, follow up periods and outcome data. Studies were classified in to 1 of 3 categories: 1) WM Training in TD children and children with ADHD; 2) Attention Training in TD children and children with ADHD; 3) Cognitive Training in children with ID.

4. Working memory training

The majority of cognitive training programs involving children have targeted the domain of WM. One intervention that has been extensively used in both TD children and children with ADHD is the Cogmed Working Memory Training program (CWMT; www.Cogmed.com). CWMT is a computerised training program that aims to improve both WM and attention problems stemming from poor WM. Each user is required to participate in multiple exercises designed to train WM every day for approximately 30 to 45 minutes. This training schedule spans over a period of five weeks and involves repeated performance on the WM exercises. Feedback and rewards are given based on the accuracy level achieved in each trial. Using the CWMT program, Klingberg, Forssberg, and Westerberg, (2002) demonstrated that a small number of children with ADHD (n=7) improved on both trained and untrained working memory tasks as a result of CWMT. In addition, training was shown to improve several non-trained skills such as nonverbal reasoning and motor activity (head movements). Similar results were found in a larger study (n=44) conducted by Klingberg and colleagues (2005) where children with ADHD were randomised to either CWMT or to a low working memory version of CWMT. In the latter version the task load remained low and did not adjust in line with the participant’s cognitive capacity, therefore lacking an adaptive component. The authors reported improvements in trained WM tasks (Span board and digit span tasks) as well as non-trained WM tasks (Visuospatial WM task; See Table 1) in the CWMT.
group. It is important to note that the CWMT program is a commercial product, as are the majority of current cognitive training programs (e.g. Brain Age, JungleMemory, Luminosity, Mindsparkle Brain Fitness Pro, WMPro), and as such the potential susceptibility to research bias as a result of conflict of interest should be considered when assessing the outcomes of commercial training programs.

Improvements seen on untrained tasks are a particularly desirable feature of cognitive training programs, and the efficacy of these programs can be assessed by the magnitude of these gains. These transfer effects can be near effects, where improvements are seen on tasks that are close to those trained. For example, improving performance on verbal WM tasks after undergoing training on visuospatial WM tasks. These near transfer effects of WM training have been consistently reported using the CWMT program (Beck, Hanson, Puffenberger, Benninger, & Benninger, 2010; Gray et al., 2012; Green et al., 2012; Klingberg et al., 2005; Klingberg et al., 2002; Mezzacappa & Buckner, 2010; Thorell et al., 2009) as well as alternative working memory training programs; Jungle Memory (Alloway et al., 2013) and Mate Marote (Goldin et al., 2013). However, some caution should be taken when interpreting near transfer effects, as the assessments used to measure improvements after training often closely resemble the tasks used in the training programs themselves. Thus improvements may simply be the result of practice effects, making it difficult to ascertain whether changes in broader cognitive skills have truly occurred. Despite the potential limitations of reported near transfer effects, the few studies that conducted follow up assessments have shown positive outcomes regarding the durability of training; with improvements being stable from 3 (Klingberg et al., 2005) to 8 months after WM training (Alloway et al., 2013).

In contrast to near transfer effects (e.g. improvements in WM), far transfer effects, where improvements occur on tasks that are quite different to those being targeted during training (e.g. attention) are far more elusive. Beck and colleagues (2010) and Green and colleagues (2012) conducted randomised clinical trials of CWMT in individuals with ADHD. Both studies reported near transfer effects (improvements in WM), however no improvements were seen in parent rated reports of hyperactivity/impulsivity or oppositional defiant behaviours (Beck et al., 2010), or ADHD symptoms (Green et al., 2012). The lack of far transfer effects suggest that WM training may only be reliable in producing changes in targeted domains and highlights the potential limits of this intervention in producing more widespread improvements. These findings are supported by additional investigations of far transfer effects as a result of WM training which have found no improvements in attention (Green et al., 2012; Thorell et al., 2009), ADHD symptoms (Green et al., 2012), response inhibition (Johnstone et al., 2011), IQ (Holmes et al., 2010) or academic achievement (Gray et al., 2012; Holmes,
Gathercole, & Dunning, 2009). However an equal number of studies have found contradictory far transfer effects after WM training, with improvements being present in untrained domains such as attention (Beck et al., 2010; Goldin et al., 2013), ADHD symptoms (Johnstone et al., 2011; Klingberg et al., 2005; Mezzacappa & Buckner, 2010), response inhibition and reasoning (Klingberg et al., 2005; Klingberg et al., 2002).

Collectively these findings suggest that WM training is inconsistent in producing improvements in untrained domains, and emphasizes potential constraints of training programs that seek to improve a wide range of cognitive skills. However it is also possible that the variance in results can be attributed to the assessment of transfer effects. For example, several studies have investigated transfer effects of WM training to inhibitory control, with some showing clear improvements in inhibition (Klingberg et al., 2005; Klingberg et al., 2002), while others show no improvements (Thorell et al., 2009). Although the same task was used (Stroop Task), the measures of performance varied significantly across the studies, with differences in task administration and importantly outcome variables (e.g. error rates and reaction times). These findings highlight the difficulties surrounding the assessment of WM training, and draw attention to limitations which likely contribute to the irregularities in reported training effects (such limitations are discussed in detail later in this review).

Insert Table 1 about here

4. Attention training

Given that the core feature of ADHD is the large magnitude of attention deficits, it is unsurprising that the majority of attention training programs have targeted this clinical population. One of the first studies to examine whether attention training was beneficial for individuals with ADHD used a non-computerized program called ‘Pay Attention!’ (Kerns, Eso, & Thomson, 1999). This program targets several components of attention (selective attention, sustained attention, alternating attention and divided attention) via a series of activities using illustrated playing cards. Children allocated to the treatment group carried out tasks such as sorting the cards in stacks based on the card characters gender, or turning over the cards where characters were wearing a hat. In contrast, the comparison group was given a variety of academic oriented computer based puzzles and games (see Table 2). The results showed that improvements in sustained, selective and executive attention did occur as a result of training using the ‘Pay Attention!’ program, most notably on tasks measuring selective attention. In addition, improvements were also seen in non-trained skills such as mathematical competence. These results provide promising support for the potential of attention training programs to produce
both near and far transfer effects. However it is important to note that although an active control group was included, the medium used to deliver the intervention and control programs differed substantially, with one being computerised (control group) and the other non-computerised (intervention group). These disparities raise several concerns regarding the ability to accurately compare the performance of children in each of these groups, particularly when the majority of outcome measures were non computerised tasks. In addition despite improvements on objective measures after training no differences in parent reports of inattention or hyperactivity were observed in either the intervention or control group. It is possible that the discrepancy in parent rated reports is due to expectancy bias, as parents in both groups reported lower levels of inattentive behaviour after training. These findings highlight the need to include both objective and subjective outcome measures when evaluating training programs to prevent participant bias masking potential training effects.

Further studies using the ‘Pay Attention!’ program with larger samples have shown training-induced improvements in attention in children with ADHD as well as improvements in parent reported inattentive behavior (Tamm, Epstein, Peugh, Nakonezny, & Hughes, 2013). For example, Tamm et al. (2013) reported significant improvements in attentional processes including selective, sustained, divided and switching attention after an 8 week training period when compared to no training. In addition, parent and clinical ratings of ADHD symptoms and child self-reports of ability to focus showed improvements after training. Similar positive effects have been shown using computerized attention training programs in children with ADHD (Shalev, Tsal, & Mevorach, 2007). Using a randomized control trial, Shalev and colleagues (2007) assessed a computerized attention training program on 36 children with a diagnosis of ADHD. This training program consisted of four sets of tasks that targeted sustained attention, selective attention, orienting of attention and executive attention. Participants were either assigned to the training program or to an active control group; both groups participated in the same number of sessions, however, individuals in the control condition played computer games and various paper and pencil activities during their sessions. These computer games contained automatic scoring and feedback as well as multiple levels of difficulty. The results indicated that participants within the intervention group showed improvements in parent reports on inattention; these effects were not present in the control group. However as in Kerns et al’s (1999) study, the platform used to deliver the intervention differed from that used in the control group, with the active control group engaging in non-computerised tasks, as well as adaptive computerised tasks. The nature of the computer games are not disclosed in the article, making it hard to ascertain whether these tasks could have potentially confounded the results.
Despite the outlined limitations the positive findings of computerised attention training programs in ADHD have been mirrored in studies of TD children, where more robust control tasks (only computerised) have been employed (Rabiner, Murray, Skinner, & Malone, 2010). However it should be noted that both of these computerised training studies (Rabiner et al., 2010; Shalev et al., 2007) only assessed changes in behavioural attention, and did not include objective measures to assess improvements in cognitive attention (see Table 2). It is possible that improvements in behaviour may have occurred due to a number of other factors, such as simply adhering to a structured training schedule and listening to instructions over an extended period of time. The lack of objective measures makes it difficult to attribute behavioural improvements to changes in underlying cognitive processes and neural structures associated with the targeted training domain. Indeed, when long term follow ups have occurred the reported behavioural improvements have not been shown to be stable a year after training has ceased (Rabiner et al., 2010). This lack of stability may indicate that the core cognitive processes that drive these behaviours were not improved as a result of training. Alternatively it is possible that like physical exercise, cognitive exercise needs to be ongoing for sustainable benefits to occur.

In parallel to the inconsistencies observed in WM training studies regarding transfer effects, attention training studies have equally shown disparity in their findings. Although near transfer effects are evident across both computerised (Rabiner et al., 2010; Shalev et al., 2007) and non-computerised attention training programs (Halperin et al., 2012; Kerns et al., 1999; Tamm et al., 2013), there is contradictory evidence regarding far transfer effects. For example some attention training programs report improvements in executive functioning (Steiner, Sheldrick, Gotthelf, & Perrin, 2011; Tamm et al., 2013), WM (Kray, Karbach, Haenig, & Freitag, 2012), reading comprehension (Shalev et al., 2007), academic achievement (mathematics) and planning (Kerns et al., 1999). Although these effects seem promising, several studies present evidence demonstrating that attention training does not produce improvements in WM (Kerns et al., 1999; Tamm et al., 2013; Wass, Porayska-Pomsta, & Johnson, 2011), reasoning (Kray et al., 2012), academic achievement (Rabiner et al., 2010) or inhibition (Tamm et al., 2013). These findings alongside those reviewed in WM training programs underline the potential constraints of this approach in producing broad cognitive and behavioural improvements.

4.1 Executive Attention Training

One specific area of attention that has been highlighted as being particularly susceptible to cognitive change is executive attention. Executive attention involves the monitoring and resolution of conflict through efficient control of a range of attentional mechanisms (i.e. shifting, disengaging, and alternating attention). This network has several overlaps with both attention and working memory, and encompasses a number of
higher order skills associated with cognitive control, for example inhibition, set shifting and planning (Rueda, Posner & Rothbart, 2005a). Due to its complex nature this network is relatively slow to develop and runs in parallel to the drawn out maturation of its associated brain region, the prefrontal cortex (Toga, Thompson, & Sowell, 2006). The gradual refinement of this network means that there is a considerable period of time when developmental change can occur and thus emphasises the potential for environmental modification. A particularly comprehensive study that assessed executive attention across a wide age range of children (4-13 years), demonstrated that the main weakness in younger children was cognitive flexibility which required continually switching back and forth between rules (Davidson et al., 2006). Kray and colleagues (2012) aimed to improve this area of weakness in children with ADHD via a task switching training program. Children were randomly assigned to one of two groups; one group started with a single-task training program and then performed a task-switching training program and the other group vice versa. The single task training sessions consisted of either a ‘transportation task’ whereby children had to decide whether a picture displayed a car or a plane; or a ‘number’ task where children had to decide whether there was one or two planes/cars presented. In the task-switching training session’s participants had to switch between both of these tasks. The results of the study showed that the task-switching training lead to improvements in task-switching performance, whereas the single task training program did not. Additional improvements were seen in verbal WM and inhibitory control, which is a process encompassed within executive attention (Kray et al., 2012).

Further support for the potential of executive attention training programs is provided by Rueda and colleagues (2005b), who investigated its effects in TD children. Children were invited to participate in a training program that covered 4 main areas of executive attention: anticipation / tracking, attention focusing / discrimination, conflict resolution, and inhibitory control. Improvements in executive attention were found, with both 4 and 6 year olds showing improvements in resolving conflict after training, however these improvements did not differ statistically from those observed in the control group. Although improvements in executive attention measured via cognitive assessments could not be attributed to intervention effects, electrophysiological data suggested that training influenced the scalp distribution of event related potentials (ERPs). Specifically, 4 year olds in the training group produced an EEG pattern in the prefrontal region that was similar to untrained 6 year olds, whereas 6 year olds in the training group produced more adult-like patterns (Rueda et al., 2005b). Equally impressive findings were shown in a recent study which demonstrated that 5 year old TD children activated executive attention networks within the brain quicker and more efficiently than controls after only ten sessions of attention training (Rueda, Checa, & Combita, 2012). Of critical importance is
the fact that these effects were present up to 2 months after training. Taken together, these studies show that the executive attention network alongside its associated brain systems are susceptible to environmental change, and implicate this domain as a potential focus for subsequent training programs.

However it should be noted that not all executive attention training programs have produced such promising effects. For instance training programs that have focused on improving inhibitory control have shown limited results in both children with ADHD, and TD children respectively (Johnstone et al., 2011; Thorell et al., 2009). Thorell and colleagues (2009) reported that the lack of improvement in inhibitory control may be due to difficulties in making this type of training program adaptive. Unlike WM training programs where difficulty can be increased easily by simply adding more items to recall or by expanding the amount of time that information needs to be retained for, much less is known about manipulating the difficulty of inhibitory control tasks. This adaptive component is therefore critical in obtaining gains from cognitive training, and may help to distinguish which domains are suitable targets for this type of intervention.

Insert Table 2 about here

5. Potentials of Cognitive Training for Children with Intellectual Disabilities

Although EF difficulties are present in children without ID, such as those with ADHD, it is the severity of these difficulties and the subsequent impact on cognitive development that distinguishes them from individuals with EF difficulties derived from a specific neurological basis. Unlike children with ADHD who are primarily referred to interventions for their behavioural problems (Pelham, Fabiano, & Massetti, 2005), those with ID have a wide range of cognitive weaknesses alongside behavioural features that require treatment. In particular, attention difficulties are highly prevalent in children with ID’s such as Down syndrome (Capone, Goyal, Ares, & Lannigan, 2006; Cooper et al., 2009), Williams Syndrome (Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006), Fragile X syndrome (Bailey, Raspa, Olmsted, & Holiday, 2008) and Autism (Elsabbagh, et al., 2011). In many cases children with ID share similar behavioural attention profiles, characterized by poor inhibition, problems concentrating and impulsivity. Indeed the prevalence of ADHD among children with Down syndrome (DS), the most common genetic cause of ID has been shown to be very high, with current estimates reaching 43.9% (Ekstein, Glick, Weill, Kay, & Berger, 2011). Equally Williams Syndrome (WS) another genetic disorder has significant comorbidity with ADHD (Rhodes, Rihy, Matthews & Coghill, 2011); 65% of children aged between 4-16 years meet the criteria for ADHD (Leyfer et al., 2006).
Despite the commonalities in observable/behavioural attention deficits across ID, it is likely that the cognitive pathways that have led to these behavioural end points are unique to each specific disorder. It is these unique underlying cognitive profiles in children with ID that may benefit the most from tailored cognitive training programs that are able to target specific areas of weakness. Indeed cognitive training is thought to be most effective when used in individuals who have profound weaknesses in the area being trained, that is individuals with lower initial performance levels often benefit most from training (e.g. Karbach & Kray, 2008; Rabiner et al., 2010). Despite this there are currently very few training programs that target children with reduced cognitive capacity and therefore the extent to which training-induced improvements may occur in children with severe EF difficulties is presently unknown.

5.1 Attention Training vs Working Memory Training in Intellectual disabilities

One of the few studies that has assessed the efficacy of cognitive training in children with ID, examined the feasibility of the CWMT program (Soderqvist et al., 2012). Forty one children with ID (defined as an IQ below 70) were randomly allocated to either an adaptive or non-adaptive version of the CWMT program. After a 5 week training period involving purely visuo-spatial tasks, improvements in tasks assessing verbal WM and language functions were observed in the adaptive training group. The authors note that these findings are of particular importance for those with ID, as verbal WM deficits are often shown to be more severe than visuo-spatial deficits (Van der Molen, et al., 2009), thus highlighting the potential to produce improvements in areas of relative weakness. However the authors also highlight that the effects of the treatment were shown to vary significantly between children, with some children showing little if any progress at all during training. These individuals did not progress considerably above the levels of the non-adaptive training program, indicating that some individuals in the intervention group were actually subject to the same training conditions as those in non-adaptive group. The lack of a control group poses significant difficulties when attempting to compare progress across the groups and prevents the reported improvements being unequivocally associated to the adaptive WM training program. In addition to these methodological weaknesses, no training effects were observed a year after training. However this study is useful in highlighting that intensive computerized cognitive training is feasible in children with ID and provides evidence regarding predictors of training success within this population. The strongest predictors for progress were gender, comorbidity (an additional diagnosis alongside ID e.g. neurological disorders, epilepsy) and baseline capacity on verbal WM. The latter is particularly important as individuals who had higher verbal WM at baseline achieved the most amount of progress during training and a higher level of transfer effects, suggesting that a minimum WM
capacity may be required to produce training related improvements. This is of particular importance when assessing whether cognitive training is appropriate for individuals with ID, as processes such as WM are already severely compromised and may prevent children from engaging with the program at all. Indeed children with Autism have been shown to have impairments in WM (Steele, Minshew, Luna, & Sweeney, 2007) as well as those with WS, (Menghini, Addona, Costanzo, & Vicari, 2010; Rhodes, Riby, Fraser & Campbell, 2011), FXS (Munir, Cornish, & Wilding, 2000; Schapiro et al., 1995) and DS (Lanfranchi, Cornoldi, & Vianello, 2004).

These weaknesses in WM were highlighted by Bennett, Holmes and Buckley (2013) who attempted to use the CWMT training program in 25 children with DS between the ages of 7-12 years. Children were allocated to either the intervention group or a waiting list control group. An initial pilot indicated that children found the majority of tasks in the CWMT program too difficult and that the training session of approximately 30 minutes was actually taking 50 minutes. These findings highlight a crucial limitation of current training programs which overlook the unique profiles of children with ID and fail to account for reduced cognitive capacities. Although most training programs are adaptive and involve incremental increases in task difficulty, the basal levels at which the programs commence are often too complex for those with ID. In order to account for this, Bennett and colleagues (2013) used an alternative training program that was designed for preschool children, the Junior Cogmed Working Memory Training (JCWMT) program, which involves visuospatial WM tasks. After WM training, the intervention group showed significant improvements in both trained and non-trained visuospatial short term memory tasks (see Table 1). These improvements in non-trained memory skills were shown to be sustained for 4 months after training had ceased. Correspondingly parent reports on behavioural measures of EF also showed improvements in the intervention group after training. These findings provide the first evidence that cognitive training can produce significant and stable improvements in both cognitive and behavioural EF in children with ID. However some inconsistency in near transfer effects were seen, as no improvements were present in verbal WM skills. As suggested by Soderqvist et al (2013) it is possible that existing weaknesses within the cognitive profile of those with DS may have attributed to these discrepancies. For instance, individuals with DS have been shown to have more impairments in verbal WM than visuospatial WM tasks when compared to TD controls (Lanfranchi, Cornoldi & Vianello, 2004).

Considering the evidence that verbal WM is impaired in individuals with DS, baseline performance on verbal WM tasks may be an indicator of susceptibility to training induced plasticity. In contrast to Bennett et al (2013), Van der Molen and colleagues (2010) who allocated 95 individuals with mild to borderline ID (IQ: 55-85) to one of three conditions (adaptive WM program; non-adaptive WM program; control program) managed
to produce improvements in verbal WM but not visuospatial WM. These converse findings highlight the importance of considering the unique and diverse cognitive profiles of individuals with ID when attempting to produce cognitive change and indicate that participants with low levels of plasticity may require alternative methods of training, such as altered durations or more tailored tasks.

The inconsistencies in near transfer effects in training studies involving children with ID are further compounded by a lack of far transfer effects; with no improvements reported in reasoning skills (Soderqvist et al., 2012) or inhibitory control after training (Bennett et al., 2013; Van der Molen et al., 2010). The lack of far transfer effects as well as the variability in training success in children with ID may indicate that WM is not the most appropriate domain to target in order to produce extensive and stable cognitive change. Instead other domains which have a greater influence on overall cognitive control should be considered. It is possible that attention may be a more apt training domain, predominately because attention skills have been shown to emerge earlier in development and prior to the emergence of WM skills (Gazzaley, Cooney, Rissman, & D’Esposito, 2005; Gazzaley & Nobre, 2012; Rutman, Clapp, Chadick, & Gazzaley, 2010). As previously mentioned, cognitive training has been shown to be most effective when implemented early in development (Wass et al., 2012), and it seems that attention training programs are able to facilitate this early intervention and target a younger age bracket than WM training programs. Interestingly when comparing participants in WM training studies to those in attention training studies, there is a stark contrast in the ages of participants, with those in the attention programs being notably younger (see Tables 1 and 2). Particularly in children with ID, where the knock on effect of EF difficulties is so great, the ability to target these core skills at a young age is imperative.

Further support for the assertion that attention may be a more beneficial training domain for young children is shown through investigations of both WM and attention training in infancy (Wass et al., 2011). Although this investigation examined TD infants (11 months old), the results are still informative for children with ID as neuronal development is often delayed within this population and cognitive abilities are more likely to resemble that of children much younger than their own chronological age. In this study infants attended 5 lab based sessions across a period of 15 days, during each session infants had to take part in 4 tasks, which targeted: sustained attention, inhibition, selective attention and visuospatial WM. The authors found that even with this relatively short training period improvements were seen in sustained attention, saccadic reaction times (e.g time taken to look at a stimuli) and the ability to disengage visual attention (e.g. the ability to switch gaze from one stimuli to the next). However WM was not shown to improve as a result of training, suggesting that WM training may only be effective later in development when this function has begun to emerge. In children with
ID, where difficulties in WM and attention are both present it is likely that the lack of cognitive foundations initially laid by attention skills may impact significantly on the development of more complex skills such as WM. Although theoretical evidence suggests that attention training programs may be highly beneficial to children with ID, to date no studies have investigated this possibility. The lack of research in this area makes it difficult to establish the full potential of this intervention and prevents clarification of which domains are likely to produce the most promising results.

However what is known, is that the attention profiles of children with ID are unique, and that specific strengths and weaknesses are present across the core attentional networks. For example individuals with DS and WS have shown specific weaknesses in selective attention and executive attention (Breckenridge, Bradick, Anker, Woodhouse, & Atkinson, 2013a). Whereas sustained attention has been shown to be a relative strength in DS (Breckenridge et al., 2013a; Cornish, Scerif, & Karmiloff-Smith, 2007). In addition the impact that attentional constraints have on specific aspects of development in children with ID, has been shown to be syndrome specific, with ADHD like behaviours predicting poor literacy skills in children with DS, but not those with WS (Cornish, Steele, Monteiro, Karmiloff-Smith & Scerif, 2012). Equally underlying weaknesses in attention have been shown to influence behavioural features of ID. For instance, the tendency to ‘over focus’ on particular stimuli in autism (Liss, Saulnier, Kinsbourne, & Kinsbourne, 2006; Patten & Watson, 2011), as well as the repetitive and stereotyped behaviours which are commonly reported in autism, are suggestive of impairments in inhibitory control (Richler, Huerta, Bishop, & Lord, 2010; Rodgers, Riby, Janes, Connolly, & McConachie, 2012). Equally the unique behavioural profile of individuals with WS which is characterized by a tendency to indiscriminately approach both familiar and unfamiliar individuals (Jones et al., 2000), has also been attributed to weaknesses in inhibitory control (Little et al., 2013; Porter, Coltheart, & Langdon, 2007). Specifically, individuals with WS who showed the most amount of social approach to strangers also struggled the most on a response inhibition task (Little et al., 2013). Collectively these findings emphasize the strong link between underlying cognitive skills and behaviour, providing optimism that remedying cognitive weaknesses (as is the aim of cognitive training) may also produce improvements in associated behaviours. In addition it is apparent that attention profiles are not only syndrome specific but have considerable within syndrome heterogeneity. These findings indicate that interventions such as cognitive training need to be guided by research in order to ensure they are tailored to the individual needs of those with ID. For instance recent work into the development of attention in young TD children (3 to 4.5 years) has demonstrated that a two factor model of attention (sustained attention & selection and response) is more appropriate at this early stage in
development (Breckenridge, Braddock & Atkinson, 2013b). This is particularly relevant for children with ID, whose cognitive functions are likely to be more in line with younger TD children than their chronologically age matched peers. These newly defined areas of attention may therefore provide more appropriate target training domains for children with ID.

6. Discussion

The current review has evaluated to some extent the effectiveness of cognitive training programs in TD children and children with ADHD, and has also outlined the potential of these programs for children with ID. However before any firm conclusions can be drawn about the efficacy of this intervention there are several fundamental limitations regarding both the design of cognitive training programs and the studies that aim to assess them that need to be addressed.

It is apparent from the current training studies, that there are significant inconsistencies in the training regimes that are implemented (see Tables 1 and 2). Both the intensity and duration of training appears to differ substantially across studies; crucially these are factors that has been emphasized as key predictors of training success (Jaeggi, Buschkuehl, Jonides, & Perrig, 2008). Indeed the importance of training frequency and its impact on reported resulted, is highlighted by Alloway and colleagues (2013) who demonstrate that WM training benefits are greater after high frequency training when compared to low frequency training regimes (Alloway et al., 2013). Although this study did not investigate children with ID, it did examine those with learning difficulties who are likely to have distinct impairments in the targeted training domains (academic skills and working memory). Study conditions varied between, no training, low frequency training (once a week) and high frequency training (four times a week). The specific results indicated that individuals in the high frequency group improved in both verbal and visuospatial WM tasks, as well as verbal and nonverbal intelligence tests. These effects were not seen in either the low or no training conditions. These findings begin to address the significant lack of knowledge regarding optimal training frequency, and are insightful in providing evidence that training needs to be intense and repeated in order to produce stable gains.

In addition to the frequency of sessions, there is also considerable variability in the duration of training sessions, with some being as short as 15 minutes (Johnstone et al., 2011; Thorell et al., 2009), while others being over 60 minutes (Rabiner et al., 2010; Shaley et al., 2007). The average amount of time for each session tends to be somewhere in the middle of these two extremes, at around 40 minutes (Beck et al., 2010; Gray et al., 2012; Klingberg et al., 2005; Klingberg et al., 2002; Mezzacappa & Buckner, 2010; Rueda et al., 2012; Rueda et
This poses significant difficulties when attempting to train processes such as attention. It is assumed that individuals who require attention training will have existing difficulties within this domain, therefore asking them to maintain their attention for a period of 40 minutes is problematic. In particularly children with reduced attentional capacities, such as those with ID, are likely to have difficulty attending to any task for such an extensive period of time. The lack of studies that have targeted this population makes it presently impossible to establish the ideal balance between training duration and engagement with the program.

Recent studies have suggested that compliance with the program is paramount to improvements after training (Jaeggi, Buschkuehl, Shah, & Jonides, 2014). Engagement with the program is therefore vital, and although TD children may be able to rely on intrinsic motivation or financial compensation in order to complete tasks, these strategies might be ineffective in individuals with ID. Past training studies have used prizes and certificates to encourage engagement (Holmes et al., 2009), however the greatest promise comes from advances in touch interactive technologies across platforms, such as mobile phones and tablet computers. These platforms offer the potential to increase intrinsic motivation and interest in individuals with ID by incorporating elements such as immediate feedback and scoring. Cognitive training programs are able to utilise the computerised platform to ensure that the tasks included are both visually and auditorily appealing (Jaeggi, Buschkuehl, Jonides, & Shah, 2011; Klingberg et al., 2005). Importantly, new technologies are likely to appeal to those with ID by providing immediate rewards which satisfy the children’s need for rapid reinforcement (Durkin, 2010). Current evidence provides support for the potential of these platforms in children with ID, as individuals with autism have shown a preference for using technology during leisure activities (Mineo, Ziegler, Gill, & Salkin, 2009; Orsmund & Kuo, 2011) and learning activities within the classroom (Moore & Calvert, 2000; Williams, Wright, Callaghan, & Coughlan, 2002; see Van Herwegen & Riby, 2014, for a discussion on the use of technology in IDs). One of the most crucial advantages of touchscreen technology is the ability to apply these computerised interventions to very young children, this is of particular importance as early intervention has been stressed as crucial in assisting the cognitive development of young children with ID (Dawson, Rogers, Munson et al., 2010; Remington, Hastings, Kovshoff et al., 2007). Previously, the platforms used to deliver these interventions were not suitable, however the proliferation of touchscreen technologies and portable tablet computers are helping to prevent weaknesses in areas such as verbal comprehension (e.g. understanding instructions), motor skills (e.g. engaging with the interface) visual acuity (e.g. colour blindness) and hearing, from inhibiting children with ID from engaging in early interventions (Kagohara et al., 2013).
The potential benefits of computerised platforms within the area of cognitive training is further highlighted by a recent comparison of CWMT versus an alternative game based WM training program (Prins, Dovis, Ponsioen, ten Brink & van der Oord, 2011). A total of 51 children with ADHD (7 to 12 years) were randomly assigned to CWMT or a game based WM training program that incorporated clear goals and objectives within an overarching storyline. The results indicated that those who played the game based program showed greater motivation (reflected by longer training durations), better performance on the training tasks (fewer errors) and showed improvements on an untrained WM task after training (these improvements were not present in the CWMT group). These findings suggest that a computerised game format may be the most effective means to motivate and stimulate children who have difficulties maintaining attention, such as those with ADHD and ID.

Despite advances in the means of delivering cognitive training to children, there are still some limitations regarding the programs themselves. Many current cognitive training programs lack a clear underlying theoretical model, which makes it hard to ascertain which domain the programs are truly targeting. For instance, Halperin and colleagues (2012) aimed to train attention in preschool children with ADHD, however the training program used (TEAMS; Training Executive, Attention and Motor Skills) targeted an array of neurocognitive domains including inhibitory control, working memory, motor control, attention/ tracking, visuospatial abilities and planning. Although improvements were seen in ADHD symptoms, inattentive behaviour and hyperactivity, there is no way to attribute these improvements purely to 'attention' training; particularly when no pre or post training assessments were conducted for the other targeted domains (WM, inhibition, motor or planning abilities). Future studies need to establish which domains can be effectively improved via cognitive training and which cannot. Additionally, as the amount of time spent training a particular construct has been shown to be positively linked to the degree of transfer effects (Bergman Nutley et al., 2011), it is crucial that future training programs follow a more targeted approach.

Furthermore, the variety of comparison groups and the nature of the non-intervention group used in cognitive training studies also pose significant barriers when trying to establish the magnitude of training success. Surprisingly several studies have not included a control group, and have simply looked at improvements before and after cognitive training (Halperin et al., 2012; Holmes et al., 2010; Mezzacappa & Buckner, 2010). The implications of having no control group can dramatically influence the quality of reported training effects by inflating the amount of training benefits, and ignoring the possibility of naturally occurring improvements in line with development. Despite the fact that some studies have employed a more robust design
to assess the efficacy of training such as randomized control trials, these studies often include waitlist/passive controls who do not receive any training or engage in alternative tasks (Beck et al., 2010; Bennett et al., 2013; Johnstone et al., 2011; Rabiner et al., 2010; Steiner et al., 2011; Tamm et al., 2013). Although this is preferable to no control group, it still remains difficult to assess whether observed improvements are the result of the training program, or whether they are simply the result of repetition effects or intervention effects (e.g. having to adhere to a set schedule). Future studies should ideally recruit an active control group who undertake an alternative non-adaptive intervention that has a similar training schedule to the assessed training program, as well as a passive control group. Collectively these limitations highlight the necessity for caution when gauging the impact of cognitive training programs thus far and additionally outline several key factors that require modification in order for this intervention to successfully advance.

7. Conclusions

Cognitive training programs that focus on domains such as attention and WM, have shown some promising results in TD children and children with ADHD. However the present literature provides insufficient evidence to truly evaluate the efficacy of such interventions. General concerns regard the need for researchers to pinpoint optimal training frequency and duration, develop theoretically driven training programs, implement studies that measure transfer effects with valid tasks, and eliminate the use of inadequate control groups. Until these limitations are addressed the magnitude of cognitive training effects cannot be reliably evaluated.

Despite the current limitations of cognitive training programs, these paradigms may provide an alternative to pharmaceutical interventions in individuals who are ‘at risk’ or already vulnerable to EF deficits, such as those with ID. This review highlights that these programs are worthy of further investigation and provides some suggestions for future research. Primarily the development of chronological and developmental age sensitive training programs that target appropriate EF constructs are imperative. In children with ID shortfalls in EF are further confounded by the significant global deficits in overall cognitive skills. For this reason, unlike children with other causes for their EF difficulties such as ADHD, for which EF training programs have proven valuable, utilising these same programs for children with ID are likely to be ineffective because they require requisite cognitive skills that are already severely compromised. These findings underscore the need to develop training programs that recognise both fundamental weaknesses but also relative strengths of children with ID. For instance although improvements in attention may occur after WM training
and vice versa; improvements in WM may occur after attention training, the latter strategy is preferable in children with ID due to an already severely reduced WM capacity.

Cognitive training constitutes a lucrative industry as evidenced by the proliferation of several commercial programs and at times the publicity surrounding this intervention has surpassed the current scientific evidence. However the positive aspects of commercialisation, such as public accessibility and funding is beneficial in prompting and facilitating scientific investigation to address the current weaknesses surrounding this intervention. In conclusion, the influx of technological advances alongside the considerable plasticity of the developing brain offers hope for improving EF in children with intellectual disabilities via training programs that integrate novel tasks that maximize ecological validity. Future studies ought to conduct well designed objective investigations of theoretically driven, tailored and adaptive training paradigms that heed the outlined limitations and take full advantage of novel emerging technologies.
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References


### Table 1.

*Characteristics of Working Memory Training Studies in Children (4 to 15 Years Old)*

<table>
<thead>
<tr>
<th>Authors</th>
<th>Program</th>
<th>Sample (Age)</th>
<th>Profile</th>
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<th>Total Sessions</th>
<th>Total weeks</th>
<th>Session time (minutes)</th>
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<th>p</th>
<th>Far Transfer</th>
<th>p</th>
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**Note:** The total sample includes participants in both the intervention and control groups who were included in analysis. Participants who did not complete the trial are not included in the total sample. The p value displayed indicates improvements in the intervention group from pre to post-memory training. 
LD: Learning Difficulties; NS: Not Significant; WM: Working Memory; CWMT: Cogmed Working Memory Training; ADHD: Attention Deficit Hyperactivity Disorder; JCWMT: Junior Cogmed Working Memory Training; DS: Down syndrome; STM: Short Term Memory; SN: Special Needs; TD: Typically Developing; RT: Reaction Time; M-BID: Mild- Borderline Intellectual Disability.

1. Dahlin (2011) implemented a passive control group for the pre training assessments but did not conduct any further assessments on this group, thus preventing any comparisons regarding training effects.

2. The authors state improvements in WM, however these are based on improvements within the game itself, and therefore do not constitute transfer effects.

3. Although improvements were seen in planning skills in the intervention group (p=.01) the same improvements were seen in the control group (p=.01) and therefore cannot be exclusively attributed to WM training.

4. Despite and overall group effect being shown for visuospatial WM, no post hoc tests are presented, nor are the differences in scores from pre to post training.
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* WM: Working Memory

** Note: The table presents studies focusing on attention training in children aged 11 months to 12 years old. The studies include various programs and control groups, with outcomes measured in terms of parent, teacher, and executive ratings. The table also includes session times, total weeks, and sample sizes. The p-values indicate statistical significance of the findings.
Note: The total sample includes participants in both the intervention and control groups who were included in analysis. Participants who did not complete the trial are not included in the total sample. The p value displayed indicates the level of improvement in the intervention group from pre to post-attention training. TEAMS: Training Executive, Attention and Motor Skills; ADHD: Attention Deficit Hyperactivity Disorder; NS: Not Significant; WM: Working Memory; TD: Typically Developing; ERPs: Event Related Potential’s; CPAT: Computerised Progressive Attentional Training Program; EF: Executive Functions.

*Rabiner et al (2010), included two intervention groups, attention training (Braintrain) and academic training (Riverdeep). The results are reported for the attention intervention.

*The age of participants is not specified. Participants are first grade students from the USA, based on this information students are presumed to be 6yrs of age.

*This study compared ERP waveforms during an attention task pre and post training, as such specific p values are not available but the authors present a series of useful topographic maps.

*Steiner et al (2011) had two intervention groups, attention training only, and attention training with neurofeedback. The results reported are for both of these attention training programs.