

Introduction

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Cognitive rehabilitation is broadly defined as a systematic intervention designed to compensate for or ameliorate the impact of cognitive and/or behavioral difficulties following a neurologic injury or illness with the ultimate goals of maximizing safety and improving daily functioning, independence, and quality of life.¹ Intervention can involve cognitive, behavioral, or pharmacologic methods. Cognitive rehabilitation is typically delivered by specialists from a variety of disciplines, including speech pathologists, occupational therapists, psychologists, and neuropsychologists. These specialists provide essential input into the design and delivery of interventions. Additionally, cognitive rehabilitation interventions for children are routinely implemented by teachers and therapists as part of the child's educational program. This book is designed as a resource for professionals of all disciplines who are interested in better understanding the existing evidence base for cognitive rehabilitation interventions for children with neurological disorders.

The inspiration for this book was our 2014 continuing education seminar at the annual meeting of the American Academy of Clinical Neuropsychology (AACN), titled, "Cognitive Rehabilitation in Children and Adolescents with Acquired Brain Injury." The focus of that seminar was evidence-based interventions in traditional acquired brain injury (ABI) rehabilitation populations, such as traumatic brain injuries, strokes, and tumors. However, the literature describing the efficacy of cognitive rehabilitation in children has also focused on conditions such as autism, learning disabilities, and attention-deficit hyperactivity disorder in addition to ABI. In fact, the term "neurodevelopmental disabilities" (NDD) is increasingly used in place of or synonymously with "developmental disabilities," consistent with an increased understanding that many childhood disorders have neurological bases.² For the purpose of this book, a neurological condition is an insult that disrupts the trajectory of brain function at any time in the course of neurodevelopment. Thus, professionals providing cognitive rehabilitation to children and adolescents with neurological conditions need to have a strong understanding of both typical and disrupted neurodevelopment.

Over the past two decades, the field of cognitive rehabilitation has expanded significantly. In adults with traumatic brain injuries and strokes, the literature on the efficacy of cognitive rehabilitation includes a growing number of studies (for review, see ^{3,4}). A manual for clinicians has also been developed that describes many of the evidence-based interventions in detail.¹ In contrast, only a few studies have examined efficacy of intervention techniques for children with brain injury or other types of neurological disorders⁵⁻⁷ and most recent reviews have focused primarily on working memory training. Although the vast majority of research has focused primarily upon adults, researchers and clinicians have begun to use

cognitive rehabilitation interventions for children and adolescents with a range of neurological conditions. However, existing evidence has not yet been comprehensively assembled into a published book.

Studies describing the efficacy of cognitive rehabilitation interventions in children with ABI and NDD as well as adults with ABI are reviewed throughout the following chapters. Reviews of cognitive intervention in children with neurological disorders highlight the absence of randomized controlled studies and the need to continue exploring the efficacy of cognitive rehabilitation in children with ABI, as well as other neurological conditions. Despite the need for more research on pediatric cognitive rehabilitation, each year the literature is growing.

In Section 1 of the book, the evidence-based literature describing the efficacy of cognitive rehabilitation interventions for several core domains of functioning is reviewed, including attention, memory, perception, language, and executive functioning. The current state of evidence-based data in the increasingly popular market known as “brain training” is also discussed in a chapter on attention/working memory (Chapter 1). Additionally, chapters focusing on particular interventions including family-based intervention and pharmacological interventions are included. In Section 2 of the book, areas of special consideration including cognitive rehabilitation issues related to school programming, transition to adulthood, advocacy, and billing practices are highlighted. Case examples are included to provide clinicians with real-world models of evidence-based interventions and practices, drawn from the extensive and personal experiences of experts in the field of pediatric cognitive rehabilitation.

Each chapter focuses on the studies with the strongest evidence. As a result, some chapters focus on a range of populations, while others primarily highlight a specific population. For example, the language chapter focuses heavily on interventions for children with autistic spectrum disorder (Chapter 6). Interestingly, while there is strong evidence to support the use of language therapy to address acquired aphasia in adults following stroke,⁸ there is virtually no literature on interventions for aphasia in children. The family-based intervention chapter focuses primarily on literature involving children with traumatic brain injuries, as there are a series of well-designed studies involving this population (Chapter 5). The two evidence-based cognitive rehabilitation chapters with the least amount of evidence are the chapters on memory (Chapter 2) and hemispatial neglect (Chapter 3). The authors of these chapters draw from a range of case studies and small case series as well as the adult ABI literature to provide a background for the reader. Although pharmacological agents are often used to promote cognitive recovery after pediatric ABI, solid evidence-based guidelines are lacking in this population (Chapter 7).

The types of interventions and underlying theoretical considerations described also vary across chapters. For instance, it has been proposed that the best context for holistic intervention is through everyday functional activities and the use of everyday people in the child’s environment. According to this theoretical model, teaching cognitive processes in an entirely decontextualized manner is not likely to be effective.⁹ However, the attention and working memory chapter (Chapter 1) includes review of interventions that primarily focus on teaching cognitive processes in this manner (e.g., computer-based adaptive programs). While the role of a coach is highlighted in some studies, others implement computer-based training alone. Although studies of massed practice are growing rapidly, these interventions can have limited generalizability to the real world. In contrast, the chapters on family-based intervention (Chapter 5), executive functioning (Chapter 4), and school-based interventions (Chapter 8) highlight the role of everyday people in the intervention strategies reviewed. Thus, in many chapters throughout this book, authors highlight the essential roles of families

and educators in the long-term care and cognitive rehabilitation of children with neurological conditions. Examining the efficacy of these interventions is particularly challenging because complex interventions have multiple components, and it is often difficult to isolate and study the active components of any intervention.

Considering cognitive rehabilitation interventions within the context of children's neurodevelopment is crucial. Given the changes in cognitive skills and functional expectations that occur in typically developing children, effective interventions for children with neurological disorders may vary depending on age and developmental level.¹⁰ Thus, in chapters describing cognitive rehabilitation interventions, the impact of development is also addressed. For example, when addressing executive dysfunction in children with neurological disorders, scaffolding and supervision are often naturally in place to help younger children (elementary school age) establish and then implement simplified, consistent routines. In contrast, older children (middle and high-school age) may not naturally have this level of adult supervision still built into their day-to-day lives, and therefore may need more support than is typical for children of their age.

Cognitive rehabilitation in pediatric populations involves unique considerations, outlined in Section 2 of the book. First, many children receive a significant portion of their intervention services within the school system. Therefore, we have included a chapter on interventions that can be provided in academic settings for children with neurological disorders (Chapter 8). This is particularly timely, given the overwhelming supply of commercially available curricula and interventions, which may or may not be empirically supported. Second, professionals working with children need to be aware of the services and supports available to help their transition from childhood into adulthood (Chapter 9).

A primary goal of this book is to provide a comprehensive overview of evidence-based clinical practice. However, we hope to provide practical guidance on professional issues and day-to-day business practices as well, as these topics impact effective access and service delivery. For example, because service utilization is jointly shared between the medical and educational systems of care, a chapter on advocacy provides the reader with an overview of programs that have been developed to help identify children in need of services and bring together these two often discrepant service delivery models (Chapter 11). This chapter also highlights the succinct need for stronger evidence, professional awareness, and coordinated efforts to develop and improve access to evidence-based delivery models. A chapter on health-care service delivery provides an overview of day-to-day provision of and barriers to pediatric cognitive rehabilitation, such as interacting with internal health-care systems and managed care companies, billing, and reimbursement (Chapter 10).

Importantly, clinicians need to realize that service delivery models vary considerably throughout the United States and around the world. In the United States, there are specific education laws that mandate services in all public school systems for children experiencing academic impact of neurological disorders; however, the access, quality, and implementation of those supports vary considerably across states. Commercial and state insurance coverage for cognitive rehabilitation services also varies drastically across regions, states, and individual plans.

In conclusion, there is an emerging literature demonstrating the efficacy of cognitive rehabilitation for children with neurological disorders. Professionals providing these services should be familiar with the relevant evidence-based literature that informs cognitive rehabilitation practices in order to deliver appropriate interventions and advocate for the provision of services and supports at home, in the school, and in the community. In pediatric neurological populations, involvement of caregivers, parents, and educators is critical to the

development and implementation of effective cognitive rehabilitation interventions. Despite the growing body of literature examining cognitive rehabilitation of children with neurological disorders, there continues to be a great need for well-designed studies to examine the efficacy of these interventions.

Future research should emphasize collaborative, multicenter, randomized controlled trials that include an adequate number of participants with specific neurological conditions to provide sufficient power to test treatment effects. Interventions should be carefully implemented with adequate assessment of treatment fidelity, outcomes that target key components of interventions, and relevant measures of generalization. Additionally, further research is needed to better understand the specific patient characteristics, symptoms, and cognitive deficits most associated with response to intervention. With an increased evidence base, rehabilitation professionals will be able to develop research-supported best practices and clinical guidelines for cognitive rehabilitation in pediatric neurological conditions, develop accessible manualized treatment programs, and translate research protocols into effective interventions for clinicians. Given the rapid expansion of evidence-based literature examining efficacy of cognitive rehabilitation interventions in children with neurological conditions, our hope is to write the second edition of this book in ten years, with much more extensive and solid evidence.

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Section 1

Evidence-based Cognitive Rehabilitation

Chapter

1

Interventions for Attention and Working Memory

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The ability to pay attention and hold information in working memory (WM) has long been recognized as important for many cognitive tasks. Attempts to strengthen or remediate attention and WM through repetitive practice of select tasks are known by a variety of names, but they typically involve interventions designed to enhance targeted cognitive skills.¹ This chapter will first briefly explore the conceptualization of attention and WM, discuss how deficits are affected by developmental age and severity of impairment, and describe general theoretical approaches to cognitive rehabilitation. Next, meta-analytic research will be reviewed. Several common standardized interventions will then be presented. Practical and clinical applications will be briefly discussed and the chapter will end with a case example. This chapter touches on research and/or treatment recommendations for children and adolescents with a history of brain injury, attention-deficit/hyperactivity disorder (ADHD), cancer, seizures, low birth weight, malaria, and human immunodeficiency virus (HIV).

Definitions of Attention and Working Memory

The conceptualization of attention and associated neurological mechanisms has evolved over the course of the last century. In 1958, Broadbent introduced the human information-processing approach and suggested that attention involves responding to select stimuli in the environment while inhibiting responses to competing stimuli.² Subsequent information-processing models can be categorized into two broad classes: early-selection theories and late-selection theories. Early-selection theories posit perceptual suppression of non-target stimuli prior to attentional processing while late-selection theories suggest that non-target stimuli are perceived but not selected for further processing.³

Attention can be subdivided into components such as attentional orientation (directing attention to a stimulus), selective attention (focusing attention on one stimulus while ignoring another), sustained attention (maintaining attention across time), and divided attention (dividing attention across multiple stimuli).⁴ Subcomponents of attention appear to involve distinct neural substrates, and it has been theorized that attention may be strengthened through cognitive training,⁴⁻⁶ which could lead to improvements in executive functioning and possibly behavioral control.¹

The construct of attention was later expanded to include WM (working memory), composed of four processes: (1) the central executive, which holds information in short-term storage while the attentional focus shifts to other stimuli, (2) the visual-spatial sketchpad, (3) the phonological loop, which briefly stores and allows manipulation of visual and phonological material, and (4) the episodic buffer, which binds information across visual, spatial,

and verbal domains, as well as across time so that it can be stored in memory.⁷ This theory of information processing is useful as it allows for multiple levels of attentional processing. Although there are a variety of WM models,^{8,9} there is evidence that both attention and WM are domain-specific and can be either verbally or visually mediated in the pediatric population.¹⁰

For the purpose of this chapter, attention is considered to be the ability to maintain focus on a stimulus whereas WM is conceptualized as the ability to cognitively hold and manipulate information in mind.³ Although many of the theoretical models of attention do not offer clinical techniques to remediate deficits, they define the constructs of attention and WM³ and provide foundations on which rehabilitation intervention programs have been developed.

Deficits in attention and WM have a variety of congenital and acquired etiologies and occur across a spectrum of impairment. Poor attention and/or WM difficulties appear to play a significant role in ADHD,^{11,12} autism spectrum disorders,¹³ and language impairments.¹⁴ Although frontal lobes are widely considered important in attention and WM, damage to multiple brain regions such as the cerebellum, basal ganglia, or white matter pathways can also result in deficits.^{15,16} Disruption of attention and WM can range from mild to severe, and although functioning usually improves over time, difficulties may persist for months or years after an injury, and full recovery may never occur.^{3,17}

Attentional control has been hypothesized to act as a gating mechanism for the acquisition of other cognitive skills such as reading,¹⁸ language,¹⁴ and mathematical skills.¹⁹ In fact, because poor attention can impact later recall, attention deficits following brain injury are sometimes undiagnosed or miscategorized as memory deficits.³ It has been hypothesized that increased WM capacity will result in gains in skills dependent on WM.^{20,21} Research about the generalizability of WM interventions frequently refers to “near-transfer” and “far-transfer” effects.^{20,22} Essentially, near-transfer effects occur when WM performance gains generalize to tasks similar to the training task (e.g., digit span), and far-transfer effects occur when WM performance gains generalize to tasks dissimilar from training tasks (e.g., reading comprehension).²²

Age and Development

Unlike in adults, in children insults to the pediatric brain occur in the context of rapid cognitive maturation, and it is important to consider a developmental perspective when evaluating attention and WM.²³ There has been a considerable debate in the field regarding the impact of pediatric brain injury on the developing brain. Central to this debate are two theoretical perspectives: (1) “early plasticity,” which suggests that an immature brain has greater flexibility and potential for good recovery and (2) “early vulnerability,” which argues that a young brain is more susceptible to poor outcome.^{24,25} In their 2011 review, Anderson et al.²⁴ argued that these competing theories represent extremes along a “recovery continuum,” with recovery dependent on injury-related factors (e.g., severity and timing of insult), individual factors (e.g., developmental stage, cognitive status, gender), and environmental factors (e.g., family interaction, access to treatment), which together contribute to the broad spectrum of outcomes.

In terms of early plasticity, evidence suggests that functional neural networks are not fully developed at younger ages, and WM and attentional control may have more potential to be remediated earlier in development.^{26–28} There is also indication that different brain structures are used to perform the same cognitive task at various ages. For example, in

laboratory studies, adolescents and adults have been found to use the dorsolateral prefrontal cortex and parietal regions during a visual-spatial WM task, whereas children utilize the caudate nucleus and anterior insula to complete the same task.²⁹ With regard to early vulnerability, skills not yet developed at the time of neurological insult may be deficient later in development, and existing skills may lag as cognitive demands increase with age.³⁰ For instance, deficits in auditory-phonological WM may not be immediately obvious in young children but may become more problematic later on, because auditory-phonological WM becomes more dominant than visual WM as children learn to read.^{31,32} Finally, children utilize different cognitive strategies at varying developmental stages, with early injuries possibly affecting the use of emerging strategies.^{33,34} In general, this evidence supports the use of a developmental framework when clinically assessing and facilitating interventions for attention and WM, and when evaluating research on interventions for attention and WM.

Wass et al. identified 37 studies that examined whether participant age was correlated with generalization of intervention-trained attentional and/or WM skills to other cognitive functions (i.e., degree to which skills transfer).²³ Sixteen of the studies included pediatric samples, and the age range across studies was 11 months to 96 years. The authors grouped research studies into either mixed-attention studies (i.e., studies that targeted subcomponents of attention such as sustained attention and switching) or studies that explicitly trained WM. When all the studies were pooled together, a weak but significant correlation between the degree of transfer of training effects and age of participants was found, particularly when using the length of treatment as a covariate because different age groups tend to receive varying lengths of interventions. In addition, there was more transfer of training for the group of WM studies than there was for the mixed-attention studies, which could be explained by the heterogeneity of attentional subcomponents targeted in the mixed-attention group. Overall, younger individuals showed a larger degree of transfer of training. Taken together, the evidence suggests that cognitive development should be considered when planning attention and WM rehabilitation in the pediatric population.

Theoretical Approaches to Attention and Working Memory Interventions

Multiple theoretical approaches have been advocated over time to address attention and WM deficits. They can broadly be classified as (1) restorative interventions involving retraining, (2) functional adaptations involving compensatory techniques, and (3) holistic context-sensitive approaches that involve both environmental and behavioral modifications.^{3,35} In general, intervention approaches are influenced by the setting in which they are implemented. For example, patients in acute care settings often require interventions to facilitate skills related to basic functioning while patients in inpatient rehabilitation settings are more likely to require techniques focused on restoring previous skills or compensating for new impairments. Partial hospitalization allows practitioners to implement and reinforce compensatory strategies with patients while simultaneously providing them the opportunity for skill practice and implementation in the community. Outpatient interventions can involve ongoing therapy and support for independent functioning with developing skills. Holistic school-based interventions may also be appropriate for the pediatric population.³⁶ Online and computerized programs allow for home practice and also may be particularly useful for individuals who have isolation precautions during treatment due to lowered immune functioning or high contagion.³⁷

Measurement of Attention and Working Memory

When interpreting research data on the effectiveness of attention and WM interventions, it is important to acknowledge that how attention and WM are measured can affect research outcomes and subsequent conclusions. For example, simple measures of attention such as recalling a span of digits may be inadequate for the measurement of WM.³⁸ More complex measures, such as inhibiting an overlearned skill of reading while identifying ink color (i.e., the Stroop task), have also come under scrutiny because the task is sometimes used to measure selective attention and at other times used to assess WM.^{38,39} There are also concerns about the use of parent and teacher ratings because the subjective nature of rating scale and unblinded raters may result in apparent improvement due to the Hawthorne effect (i.e., alterations in behavior that occur when individuals are aware of being observed).⁴⁰ However, rating scales, which may have better ecological validity than performance-based tests, can be useful measures of attention and WM training effects in daily life.

Formation of Practice Recommendations

About 20 years ago, investigators began examining the effectiveness of attention and WM interventions in the pediatric population, and in the last decade there has been an explosion of published studies on their efficacy. Despite increased interest and improved methodology, contrasting findings are not uncommon. Comparisons between research studies can be complex, as conclusions appear to be influenced by a number of factors, such as definition and measurement of attention and WM, type and length of interventions, variety of measures used to examine effectiveness, age of participants, type of control group, and population characteristics.

Several formal systems for reviewing the literature have been developed. The American Academy of Neurology (AAN) clinical practice guidelines are one example of how strength of studies can be classified. The AAN guidelines provide a basis for evidence-based practice recommendations, and classify studies based on the type of study and strength of evidence, ranging from less robust empirical evidence (i.e., Class IV) to randomized clinical trials with masked outcomes (Class I).⁴¹ See Table 1.1. Once available research is classified, the body of research can be examined to determine if evidence suggests treatment should be considered as a Practice Option, a Practice Guideline, or a Practice Standard (see Box 1.1).⁴² At present, there is no consensus in the literature about the efficacy of cognitive rehabilitation for attention and WM in the pediatric population; however, this chapter will identify when Practice Options, Guidelines, or Standards have been recommended in specific pediatric populations based on available research.

Meta-Analyses of Attention and Working Memory Training

A number of meta-analyses have been published in recent years on the available research. In 2013, Melby-Lervåg and Hulme conducted a meta-analytic review of 23 randomized controlled trials and quasi-experimental studies in typically developing children and adults to examine WM training efficacy in healthy individuals.⁴³ The results suggested reliable short-term improvements in WM. The long-term effects appeared domain-dependent, with limited evidence for visual-spatial WM but not for verbal WM. The authors suggested variations in age and clinical trial differences impacted results, and concluded that findings do not

Table 1.1 Classification of Intervention Studies

Classification	Evidence in Representative Population
Class I	Prospective studies with randomized groups and blinded, well-defined outcome measure(s). Must include clearly delineated inclusion/exclusion criteria, independent groups, limited participant withdrawal, and balanced group characteristics.
Class II	Prospective, nonrandomized, matched cohort studies with blinded outcome measure, or randomized controlled studies with a substantial flaw.
Class III	Other controlled studies that include independent assessment of outcomes or outcomes are independently attained via objective measures.
Class IV	Research not meeting Class I to III guidelines, including uncontrolled studies, case reports, or expert opinions.

Source: Edlund et al. (2004).⁴¹

Box 1.1 Definition of Practice Recommendations

Practice Standard Evidence from at least one well-designed Class I study and other Class II or Class III studies. Effectiveness of treatment for individuals with acquired neurocognitive impairment needs to be *substantially evident*.

Practice Guideline Evidence from one or more Class I study with methodological limitations, or several well-designed Class II studies. Effectiveness of treatment for individuals with acquired neurocognitive impairment needs to be *probable*.

Practice Option Evidence from Class II or Class III studies. Effectiveness of treatment for individuals with acquired neurocognitive impairment needs to be *possible*.

From Cicerone et al. (2011)⁴²

support using WM training programs to enhance cognitive functions in typically developing individuals.

That same year, Laugenbahn et al. reviewed 34 cognitive rehabilitation studies including children and adults across 8 medical diagnostic categories affecting cognitive functions.⁴⁴ Consistent with previous reviews,^{45,46} this review supported a Practice Guideline for treating attention or memory deficits in children and adolescents who undergo treatment of brain tumors. In addition, a Practice Option for the treatment of seizure-related deficits in attention and memory was recommended.

In 2014, Robinson et al. conducted a systematic review and meta-analysis of cognitive interventions for children with neurological disorders, acquired brain injury (ABI), and neurodevelopmental disorders (i.e., ADHD).⁴⁷ They included 13 randomized controlled trial studies and determined that there was a large effect size for attention and working memory treatments. However, there were concerns about heterogeneity of results and quality of evidence, so treatment recommendations were not proposed.

Most recently, a 2016 meta-analysis of 87 pediatric studies did not show reliable improvements in the generalization of WM skills after cognitive rehabilitation.⁴⁸ The review examined whether WM training resulted in far-transfer generalization to intelligence and other cognitive domains (i.e., nonverbal ability, verbal ability, word decoding, reading comprehension, and arithmetic). All studies included control groups (either treated or

untreated), as well as pre- and post-test measures. The authors concluded that WM training produces short-term specific training effects, but does not demonstrate generalization to real-world cognitive skills.

There is a growing body of research specifically examining cognitive interventions in the pediatric ADHD population. While some individual studies have concluded that deficits of attention and WM significantly improve following massed practice exercises of attention^{49,50} and WM,^{11,51,52} meta-analyses for this population suggest that cognitive training may improve immediate attention for near-transfer effects, but is either unsupported for long-term improvement in attention and far-transfer effects⁴⁰ or has limited to no effects when using blinded raters or active control groups.⁵³ WM training alone has been shown to be less effective than multi-process approaches for the reduction of ADHD symptoms.⁵³

As with ADHD, research on attention and WM training in children with brain injuries has expanded over time. However, the body of literature remains somewhat limited. In 2005, a review of available research by Limond and Leeke identified only 11 relevant studies with a total of 54 participants.⁵⁴ Additionally, the authors were unable to draw conclusions about practice recommendations based on the available evidence. Later reviews in 2013 and 2014 also determined that there was not enough evidence to make definitive treatment recommendations.^{55,56} More recently, a 2016 meta-analysis focusing on WM interventions and brain injury included 103 adult and pediatric studies; in these studies, 1,585 of the 6,113 participants were children and adolescents with WM deficits.⁵⁷ All groups in the meta-analysis, including pediatric populations, showed improvement in WM functioning, and participants (adult and pediatric) with ABI showed a trend for higher effect size. Although research in the adult literature suggests a Practice Standard for rehabilitation of attention during post-acute recovery following brain injury⁴² and a Practice Option to provide clinician-guided computer interventions,⁴² pediatric research only suggests a Practice Guideline for rehabilitation of attention to children and adolescents in recovery from acute brain injury.⁴⁶ However, these practice recommendations are arguably based on dated reviews given the amount of research published in this area over the last decade.

Overall, meta-analytic results suggest the effectiveness of attention and WM rehabilitation in the pediatric population may be population- and domain-specific, which limits broad conclusions. Continued population-specific research, particularly Class I studies, will likely prove useful.

Cognitive Rehabilitation Programs

Although the structural and functional neuronal correlates are not yet fully understood, cognitive rehabilitation programs operate on the premise that training of attention and WM in the pediatric population may result in skill improvement.^{17,58} A variety of formal attention and WM training interventions have been developed, with many administered in the form of computer programs. Most of these interventions are adaptive, meaning the difficulty of tasks is adjusted to individual performance.^{20,59} By requiring an individual to perform repetitive tasks with WM demands somewhat higher than his or her current ability, the assumption is that practice will increase WM capacity.^{20,43,59} The level of support required by trained professionals and guardians varies by program. Several of the commercially available programs are described in this chapter, although the list is not comprehensive. See Table 1.2. There is a rapidly expanding literature base on cognitive rehabilitation programs, so only seminal or recent research will be included in this chapter.