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AN EXPLORATION OF ATTENTION IN CHILDREN WITH MYELOMENINGOCELE

DISSERTATION

Presented in Partial Fulfillment of the Requirements for
the Degree Doctor of Philosophy in the Graduate
School of The Ohio State University

By

Nancy Loss, M.A.

*****

The Ohio State University

1996

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ABSTRACT

This study examined attentional functioning in children with myelomeningocele (MM), the most severe form of spina bifida. Attention was conceptualized according to Mirsky's recent theory, which posits four specific elements of attention (i.e., focus/execute; sustain; encode; and shift). Standardized neuropsychological tests of the four attentional elements were administered to 64 children with MM and 27 of their siblings (ages 8 to 15 years). Data analyses compared the attentional functioning of the children with MM to that of their siblings, and examined whether the four attentional domains predicted behavior ratings of attention and academic achievement. Finally, the role of demographic and medical background variables as predictors of attentional functioning among children with MM was explored.

Participants completed a neuropsychological test battery that included measures of the four elements described by Mirsky. Parent ratings of attentional behavior were elicited using the Attention subscale on the Child Behavior Checklist (CBCL), and achievement was assessed using the composite standard score from the Wechsler Individual Achievement Test (WIAT) Screener. Parents completed a questionnaire of demographic information, and medical information were derived from a retrospective chart review.
The results show that the attentional functioning of children with MM varies both quantitatively and qualitatively compared to unaffected siblings. First, children with MM show impaired attention compared to their siblings. Second, individual differences in attention predict behavioral functioning and academic achievement in both children with MM and siblings, although the strength of the relationship between specific attentional elements and behavioral functioning is not consistent across the two groups. For example, focus/execute was the best predictor of ratings of attentional behavior among children with MM, whereas sustain was the best predictor among siblings. Finally, individual differences in attention among children with MM are related to both demographic and medical background variables. Specific medical variables accounting for unique variance in several of the elements of attention included the number of shunt revisions, the occurrence of shunt infections, agenesis of the corpus callosum, and oculomotor abnormalities. The findings illustrate the value of theory-driven neuropsychological analyses of attention in children with specific brain disorders.
This research is dedicated to children and families with special needs. It is also dedicated to my own special family, Keith, Benjamin, Erik and Jeffrey.
ACKNOWLEDGMENTS

I would like to thank the children and families who participated in this research project. They donated an extensive amount of personal time and energy to enhance our understanding of myelomeningocele.

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    sixth-grade children's self-reports of life events and impact and concordance with mothers.
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    children with myelomeningocele. *Progress Notes: Newsletter of the Society of Pediatric
    Psychology*, 19, 10-11.

    Verbal learning and memory in children with myelomeningocele. *Journal of Pediatric
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Rationale and Overview of Purpose

Psychology has long been interested in the role of attention in cognition and human behavior. Its earliest writings, as reviewed by Cohen (1993), are replete with psychologists grappling with this most basic of psychological functions. Wilhelm Wundt chose to dedicate the first chapter of his introductory psychology textbook to the role of attention in conscious experience (Wundt, 1912). Edward Titchener (1908) addressed the construct of attention in his writings, and proposed a two-factor theory. William James examined the multidimensional nature of attention in "The Principles of Psychology" (1890), outlining the active/passive nature of attention, the selection of subject matter, and the content of "attended-to" information. Finally, W.B. Pillsbury (1908) devoted a volume exclusively to attention, emphasizing biological aspects of attention and its relationship to working memory.

In contemporary psychology, attention maintains its prominence. In fact, the study of attention has expanded into a multifocal approach which emphasizes the cognitive components, the neuroanatomical and neurochemical substrates, developmental aspects, and clinical manifestations of attentional disorders.

Developmental neuropsychology, a specialty area within general clinical psychology, explores the relationships between brain function and behavior across the life-
span. Therefore, it seems particularly well-suited to integrate the physiological, cognitive, and behavioral aspects of the attentional construct, and is the perspective from which this project is undertaken.

**Research on Attention Deficit-Hyperactivity Disorder**

Decades of research have been conducted on attentional disorders, specifically on the behavioral disorder currently labeled, Attention-Deficit Hyperactivity Disorder (ADHD). This disorder, characterized by age inappropriate levels of inattention, impulsivity, and/or hyperactivity, is the primary reason children are referred to mental health clinics in this country (Barkley, 1990). Although ADHD is prevalent in clinical populations, children diagnosed with ADHD are a heterogeneous group. In fact, concerns about specificity have prompted researchers to explore the possibility of ADHD subtypes, or even the reclassification of ADHD into a spectrum of distinct and separate disorders.

More recently, research on ADHD has been expanded to include inquiries conducted from a neuropsychological perspective. Data are beginning to accumulate which document differences between children diagnosed with ADHD and those without. In fact, neuropsychological assessment of children with ADHD has highlighted possible neuroanatomical and functional brain differences in subgroups of these children.

**Brain Dysmorphology and Function in ADHD**

Research regarding differential brain morphology in children with attentional disturbances has been fruitful, albeit somewhat controversial. As the research has expanded, however, evidence has accrued to support distinct neurophysiological differences in children with ADHD (Ricco, Hynd, Cohen and Gonzalez, 1993; Voeller,
Indeed, studies utilizing computerized axial tomography (CT) scans and magnetic resonance imaging (MRI) have documented structural differences in children with ADHD as compared to normal control and clinical control groups. For example, Shaywitz, Shaywitz, Cohen & Rothman (1983) utilized CT scans to reveal symmetry in the frontal lobes of ADHD children. This is in contrast to normal asymmetry, in which the right frontal area is larger than the left. MRI studies confirm this finding, noting symmetry in the anterior regions for children with ADHD, specifically, a decreased width in the right frontal cortex (Hynd, Semrud-Clikeman, Lorys, Novey, & Eliopulos, 1990).

MRI data have also documented differences in the corpus callosum (CC) of children with ADHD. The CC is the fiber bundle which is critical to the transfer of information between the hemispheres of the brain. One study documented reduced size of the corpus callosum in children diagnosed with ADHD as compared to controls (Hynd, Semrud-Clikeman, Lorys, Novey, Eliopulos, & Lyytinen, 1991). Size differences were found in the area of the genu (most anterior region of the corpus callosum), the splenium (most posterior area of the corpus callosum), and in area 4 (just anterior to the splenium). These researchers hypothesize that the reduced size of the genu may contribute to frontal deficits found in children with ADHD, particularly those related to motor regulation, persistence, and inhibition. They also conjectured that the dysmorphology of the CC in the splenium, i.e., the posterior area, may be implicated in visuo-spatial processing errors which have been documented in these children. These results are confounded however, by comorbidity issues. All of the children in their sample were diagnosed with ADHD and at least one other psychiatric condition (i.e., conduct disorder; oppositional defiant disorder; mood disorder), making the unique contribution of ADHD in these findings all but impossible to discern.
Dysmorphology of the CC in children diagnosed with ADHD was also found by Semrud-Clikeman et al. (Semrud-Clikeman, Filipek, Biederman, Steingard, Kennedy, Renshaw & Bekken, 1994). Methodological rigor resulted in a sample which was diagnosed exclusively with ADHD by multiple methods. Subjects with conduct disorder, depression, anxiety, or learning disorders were excluded from the study. Furthermore, all subjects had symptoms of overactivity, and were right side dominant with regard to hand, foot, and eye preference. Significant differences were found between children with ADHD and controls. Specifically, children with ADHD had smaller CC in the area of the splenium (posterior), with differences approaching significance in the areas of the posterior midbody, and the isthmus (just anterior to the splenium). No differences were documented in the anterior regions. These findings are best understood when one recognizes the neuroanatomical networks associated with these structures. Interhemispheric connections to these areas are vast and include: the occipital and inferior temporal areas with the splenium; the somaesthetic and posterior parietal areas with the posterior midbody; and the superior temporal and posterior parietal areas with the isthmus. These researchers hypothesized that the dysmorphology found in the CC is likely due to a reduced number of fibers, rather than to gross structural abnormalities. The fewer number of fibers may underlie problems with sustained attention, in that this behavior has been anatomically linked to the parietal areas (Posner & Petersen, 1990).

Semrud-Clikeman et al. (1994) made an additional noteworthy observation. For a subset of boys in their sample, all of whom were non-responders to stimulant medication, smaller measurements in 5 of the 7 regions of the CC were found. Although the small sample size (N=5) precluded multivariate analyses of this observation, it may stimulate further investigation and assist with the delineation of ADHD subtypes.
Evidence of functional differences in the brains of persons with ADHD has also begun to accumulate. For example, adult males with childhood onset of ADHD whose offspring also have ADHD have reduced glucose utilization in the right frontal area, as evidenced by positron emission tomography (PET). Reduced glucose metabolism has also been demonstrated in left parietal, temporal, and bilateral rolandic structures (Zametkin, Nordahl, Gross, et al., 1990). Cerebral blood flow studies document decreased metabolic activity in the frontal lobes and basal ganglia, with increased metabolic activity in primary sensory and sensorimotor regions (Lou, Henriksen, Bruhn, Borner, & Neilsen, 1989).

Differences between persons with attentional deficits and those without were also found in EEG studies. Klorman (1991) reviews findings relevant to ADD with respect to event related potentials (ERPs), which are measurements of electrical activity within the brain. In particular, he comments on ERP data related to endogenous brain functions, or those elicited by psychological, rather than physical characteristics of the stimuli. Research on ERPs during cognitive tests of sustained attention have found unique differences in children with ADD. Poorer test performance is marked by reduced amplitude in a specific wave, P3b (located over the parietal area). Klorman suggests this indicates a "diminished deployment of capacity" of attentional resources (p.138). Furthermore, after administering stimulant medication to children with ADD under the same testing conditions, results indicated both an increase in P3b amplitude, and a coinciding improvement in performance on sustained attentional tasks. Mann, Lubar, Zimmerman, Miller, & Muenchen (1992) also document decreased cortical arousal in ADHD boys.

Thus, data are converging to support structural and functional differences in some children with ADHD. Evidence is most robust for neuroanatomical differences in the parietal area, hypothesized to be an integral part of the attentional system associated with
sustained attention. In addition, distinctions in the corpus callosum have been found. While these results are provocative, they remain preliminary. Additional research is needed to further elucidate these relationships.

Neuropsychological Support for ADHD Subtypes

Neuropsychological inquiry into ADHD has also begun. Preliminary results of neuropsychological studies were equivocal, (e.g., Mattes, 1980; Loge, Staton, & Beatty, 1990) attributed in part to methodological issues. The literature has been criticized for discrepant diagnostic criteria, failure to control for comorbidity, and developmentally insensitive measures (Barkley, Grodzinsky, & DuPaul, 1992).

In an extensive review of the literature, Goodyear and Hynd (1992) provide data from neuropsychological studies which support the subtyping of ADHD into ADD with (ADD/H), and ADD without (ADD/WO), hyperactivity. Performance differences between these two groups were documented in 60% of the 10 neuropsychological studies they surveyed. Children diagnosed with ADD/WO were found to have poorer automaticity of facts, a higher co-occurrence of learning disabilities, sensory-localization deficits, inconsistencies in the development of reading skills, slower rates of visual search, a higher incidence of problems with anxiety, as well as a higher incidence of maternal anxiety disorders and siblings with learning disorders. In addition, these children were diagnosed up to one year later than ADD/H children. The authors contend that the profile of children with ADD/WO is suggestive of difficulties with neurocognitive output (e.g., slow cognitive speed and dysfunctional automatization of information), and indicate a deficit in selective attention. It appears therefore, that neurobehavioral differences associated with ADD/WO implicate right posterior brain differences.
In contrast, ADD/H children have greater disturbances in behavior regulation and sustained attention. They also have a greater incidence of conduct disorder, a lower incidence of learning disabilities, consistent problems with impulsivity, and a greater likelihood of familial ADHD, hyperactivity, and substance abuse. They are diagnosed at an earlier age than children with ADD/WO, and have a more consistent response to stimulant medications, typically at higher doses. Finally, evidence suggests that ADD/H children have neuroanatomical differences in anterior brain structures (e.g., frontal lobes).

Dykman and Ackerman (1993) also examined the literature in an attempt to delineate ADHD subtypes. Utilizing data they have gathered over the past several decades they provide evidence for three distinct subtypes of ADHD: ADD/H; ADD/WO; and a third group, attention deficit disorder with hyperactivity and aggression (ADDHA). Observations of enduring disruptive behaviors, as well as neuropsychological differences (e.g., Halperin, O'Brien, et al., 1990) were used to substantiate the ADDHA classification. For example, an analysis of errors on a continuous performance task suggest that children with ADDHA commit more errors related to greater impulsivity (commissions) as compared to inattention (errors of omission). They challenge researchers to consider the ADDHA group, and call for psychophysiological research as a means of further demarcating these three subtypes.

Barkley et al., (Barkley, Grodzinsky, & DuPaul, 1992) reviewed 22 neuropsychological studies specifically associated with frontal lobe functioning in children with ADD/H. As noted above, they discovered that several tests accurately distinguish ADD/H children from normal controls, particularly those associated with response inhibition. To test the validity of ADHD subtypes, they gathered neuropsychological data on four carefully defined groups: ADD/H; ADD/WO; and LD, and normal control groups.
A commonly used neuropsychological battery of tests was given to each subject. Differences were found between the attentionally disordered children and the control groups on many of the measures, however, the battery was generally insensitive to differentiating subtypes of the disorder.

**Directions for future research on ADHD and Attention**

Recent literature on ADHD reflects the exploration of the neuropsychological underpinnings of this disorder. Preliminary findings suggest that neuropsychological inquiry into ADHD will promote a greater understanding of this behavioral disorder and may prove useful in the delineation of its subtypes. While these results are encouraging, inquiry reserved to one domain of attentional dysfunction is not sufficient to promote an overall understanding of the complexities of attentional behavior in children. Prominent researchers have argued for the need to expand research efforts beyond the ADHD population. For example, Barkley calls for a greater emphasis on the measurement of attentional constructs in "other neuropathological disorders of children" (1994, pp. 76-77). This method has been shown to be quite beneficial in elucidating attention in adults. In fact, much of the research to support current models of attention has been garnered from studies of adults with known neurological impairments (e.g., Mesulam, 1981; Posner & Peterson, 1990). Therefore, the use of this model for inquiry into the attentional functioning of children may prove equally productive.

**CHILDREN WITH SPINA BIFIDA: A Neurologically impaired population**

Spina bifida is a birth anomaly which afflicts approximately 1 in 1,000 live births (Charney, 1992). The most common of central nervous system malformations, it is
manifest by an incomplete closure of the neural tube (Varni & Wallander, 1988). Myelomeningocele (MM) is the most serious form of spina bifida and is characterized by a protrusion of the spinal cord into a sac externally located above the spinal column (Wills, 1993). Immediate surgical intervention is required after birth to prevent CNS infection and to promote the best possible outcome for the child.

In addition to spinal cord defects, MM is accompanied by other brain malformations, particularly of the brain stem and cerebellum. For example, the Arnold Chiari II malformation, almost synonymous with MM, involves five principle features (Worley et al., 1994). Most prominent of these is the caudal displacement of the cerebellar vermis, the medulla, and the lower pons. The cerebellum bulges rostrally and may sit above a hypoplastic tentorium, and the majority of persons will have caudal displacement to the extent that herniated cerebellar tissue will extend below the foramen magnum. These distortions often block the natural flow of cerebral spinal fluid (CSF) within the brain, resulting in hydrocephalus. In patients whose lateral ventricles are shunted to reduce obstructive hydrocephalus, the cerebellum may be displaced upward in an exaggerated fashion, thereby displacing other normal structures (Griebel, Oakes & Worley, 1991; Tarby, 1991).

There is a controversy as to whether the hydrocephalus is the cause, or the result, of the Chiari malformation (Tarby, 1991). Regardless, the affect of hydrocephalus on neuroanatomy has been widely studied. Research on hydrocephalus reveals that axonal degeneration, loss of myelin, compression of extracellular spaces which is believed to effect neurochemical transmission, and reduced blood flow to the frontal lobes are complications associated with this condition (Del Bigio, 1993). Additional neuroanatomical abnormalities have been demonstrated in MM including dysmorphology.
of the corpus callosum and related white matter, and disturbed neuronal migration
(Fletcher et al., 1992; Gilbert, Jones, Rorke, Chernoff & James, 1986).

Historically, infants born with MM had a very poor prognosis. Primarily due to CNS infections and hydrocephalus, the natural course of MM resulted in an 81% mortality rate within the first year of life (Lawrence & Tew, 1971). Since the 1950's, however, advances in medical technologies have significantly improved the survival rate of children born with MM. Surgical repair of the myelomeningocele within 24 hours of birth and the management of hydrocephalus via shunt placement in approximately 85% of all children with MM are the leading contributors to this improved prognosis (Wills, 1993). Although the risks associated with brain surgery have significantly decreased over the years, there remains a 5% chance that ventriculitis (brain infection) will result. Shunt malfunctions and significant brain stem involvement are other factors which contribute to poorer outcome. Current mortality rates associated with MM are approximately 18% (Steinbok, Irvine, Cochrane & Irwin, 1992).

The decrease in mortality of MM has left researchers questioning the quality of life these children will face. Physical adaptation is largely related to the location of the myelomeningocele lesion, with a higher lesion producing greater disability (Lawrence & Tew, 1971). Ninety percent (90%) of children with MM have significant urological problems and will require some form of intervention to achieve continence of urine. Orthopedic, visual and oculomotor, and cutaneous problems also accompany MM, which result in a need for close medical supervision and frequent intervention. About half of all children with MM will be able to ambulate in the community without a wheelchair, most
assisted with foot and leg braces. With age and increased body weight, however, the
majority of persons with MM become wheelchair dependent (Steinbok et al., 1992;
Wolraich & Hesz, 1988).

Psychological outcome has also been studied extensively in MM. Research
indicates that children with MM represent a highly heterogeneous population. In general,
children with MM without hydrocephalus or with shunted hydrocephalus score below
average but within the normal range on most tests of cognitive functioning. For example,
results of intelligence testing indicate that approximately 70% of children with MM have
an IQ above 80 (Hirsch, 1994). Those children without evidence of hydrocephalus have
the best cognitive outcome, and generally score 10-20 points higher than children with
shunted hydrocephalus (Wills, 1993).

The deleterious effects of hydrocephalus have been widely examined in this
population. In general, damage caused by hydrocephalus is dependent upon the rate and
magnitude of ventriculomegaly (ventricle expansion), the proximity of the structure to the
ventricle, and the developmental stage at which hydrocephalus occurs (Del Bigio, 1993).
With respect to treatment, the timing of shunt placement appears quite influential as well.
The results of one study suggest that the likelihood of an IQ of 80 or above is twice that
for children whose shunt was placed within the first 2 months of life, when compared to
those children who were shunted after age 2 years (Hirsch, 1994). While discrepant results
exist in the literature, recent trends indicate that the number of shunt revisions do not
appear to dramatically effect intellectual outcome (Steinbok et al., 1992; Wills, 1993).

Although most children with MM score within the low average range on tests of
intelligence, they tend to exhibit deficits in nonverbal skills (Hurley, Laatsch, & Dorman,
1983; Tew, 1977). In particular, large discrepancies have been noted between Verbal and
Performance IQ scores, with more robust verbal abilities noted (Dennis et al., 1981). Deficits in visual-spatial organizational tasks, attention, abstraction and behavioral adjustment have also been found (Holler, Fennel, Crosson, Boggs, & Mickle, 1994). Furthermore, although neuropsychological performance does not appear to decline with age, academic performance does (Donners, Rourke, & Canady, 1991; Wills, Holmbeck, Dillon & McLone, 1990). Researchers suggest that the pattern of impairment is similar to that described by Rourke (1987) as a nonverbal learning disability (NLD) (Wills et al., 1990; Hurley & Dorman, 1994). It has been hypothesized that NLD may be associated with damage to white matter association tracts (Rourke, 1987). Although the pattern of NLD is similar to that of children with MM, it is not synonymous. Indeed, children with MM have a differential neuropsychological profile (e.g., difficulties in linguistic ability and rote learning) as compared to children with NLD (Yeates, Enrile, & Loss, in press).

In a recent comparison of MRI imaging to performance on cognitive measures, Fletcher et al. (1992) found significant correlations between neuroanatomical differences in children with hydrocephalus (including children with MM) and their performance on cognitive tests. In this sample, a reduced size of the corpus callosum and internal capsule was significantly correlated with poor performance on nonverbal cognitive measures. In particular, damage to the corpus callosum was discovered in the posterior bodies and splenium. These areas have been implicated in visual attention, higher-order cortical function, and sensorimotor integration, as well as being the site of connection between visual areas and the occipital lobe. Speculation as to how callosal dysmorphology evolved in children with MM was provided. First, prenatal congenital abnormalities may result in partial or complete agenesis of the corpus callosum. Second, significant hydrocephalus is known to distort the corpus callosum by stretching, thinning, or destroying these white
matter structures. Thus, it seems possible that both embryologic and post-natal factors may contribute to the differences found in the corpus callosum of children with MM.

In a recent review, Wills (1993) provides possible explanations for the relatively impaired visual-motor skills seen in children with spina bifida. First, actual motoric slowing and incoordination, and poor eye-hand coordination may systematically affect performance on nonverbal tasks. Secondly, although children with MM score relatively well on verbal subtests of IQ measures, the hyperveral quality noted in these children may contribute to an overestimation of their verbal reasoning capacity. Thirdly, disturbances in visual-motor functioning may reflect an overall deficit in executive functions. In fact, Wills (1993) contends that exploration of attention, concentration, and executive functioning in children with spina bifida remains an area of "blank ignorance" (p.260).

**Exploration of Attention in children with MM**

The above information serves as a backdrop to justify further exploration into the attentional functioning of children with MM. Arguments in support of this effort are threefold. First, spina bifida is the most common CNS birth defect. In the last few decades survival rates have increased dramatically within this population. Swift and effective management of accompanying hydrocephalus has resulted in an improved cognitive outcome. Although more children with MM function within the normal intellectual range, many of these children evidence learning differences. Inquiry into the nature of these differences should serve to maximize the learning potential of these children. Second, systematic exploration of attentional functioning in this population is lacking, even though clinical observation suggests that attention is impaired in this group of children. Furthermore, the potential contribution of attentional dysfunction to the known cognitive
deficits in this population have not been adequately explored. Third, neuropsychological inquiries into ADHD have challenged researchers to compare attentional functioning in children with ADHD to that of children with known neuropathology. It is expected that this method of inquiry will help to elucidate attentional functioning in children, much as it has for the adult population.

To provide theoretical direction to the present inquiry, Mirsky's model of attention is used. It is chosen for several reasons. It is the only model of attention currently available which has been empirically developed utilizing both child and adult samples, and using standardized neuropsychological tests. In addition, the components proposed by Mirsky seem relevant to the questions which exist in the current literature. For example, Wills (1993) states that the deficits seen in children with MM may reflect underlying problems with attention. Specifically, she suggests that these children may have problems with tracking, focusing, selection, sustaining, and flexibly shifting set as needed. Mirsky's model addresses most of these issues.

Mirsky's Taxonomy of Attention

Mirsky (Mirsky, Anthony, Duncan, Ahearn, & Kellam, 1991; Mirsky, Lochhead, Jones, Kugelmass, Walsh, & Kendler, 1992) initially proposed a multicomponent taxonomy of attention which emphasized three elements: focus, sustain, and shift. The "focus" element represents the ability to select relevant target stimuli from a broad array for further processing. "Sustain", is defined in terms of vigilance, or the capacity to maintain focus and alertness over time. "Shift" is the ability to change the focus of attention in a flexible and adaptive manner.
To test his model, Mirsky et al. (1992) analyzed data from two large samples: an adult sample of clinical neuropsychiatric patients and normal controls (N=203); and a sample of children (N=435) from a metropolitan public school system with no known neurological problems. Multiple standardized neuropsychological measures of attention were given to each subject. When analyzed, data from each sample loaded on four factors instead of the hypothesized three. These factors were labeled: focus-execute; sustain; shift; and encode. Even more recently, based on collaborative efforts (Tatman, 1992; Tatman, Fantie & Mirsky, 1995; as cited in Mirsky, 1996) Mirsky has entertained a fifth component of attention, "stabilize", which refers to the reliability of attentional effort (Mirsky, 1996).

Mirsky has also delineated neuroanatomical substrates associated with these functions. The focus-execute function captures behaviors associated with visual scanning and rapid verbal or motor response. It is believed to be mediated by three regions of the brain: the inferior parietal, superior temporal, and striatal regions. The posterior parietal cortex is an area of convergence for all motor regions, as well as sensory, limbic (i.e., cingulate cortex), thalamic, and reticular areas of the brain stem. It is also connected with the corpus striatum, specifically the caudate nucleus which is contained therein. Functionally associated with focused attention, the superior temporal sulcus integrates multilmodal sensory information. The complexities represented by these neurosubstrates is reflected in the behavioral characteristics of focus-execute. This factor requires the ability to translate sensory information into complex motor responses in relationship to given task requirements.

The sustain factor is believed to be mediated by the most primitive regions of the brain. Specifically, Mirsky proposes that it is associated with the brain stem (i.e., tectum and mesopontine areas of the reticular formation) and midline and reticular thalamic
nuclei. He cites research by Bakay Pragay et al. (1978) which "identified cells in the midline thalamus, deep layers of the superior colliculus, tectal and pretectal regions, and pontine and mesencephalic portions of the brain stem" with sustained attention.

Functionally, sustained attention is associated with the ability to maintain consciousness and appropriate levels of arousal. Mirsky believes these functions represent one of the most phylogenetically primitive attentional abilities.

The prefrontal cortex is believed to be responsible for the capacity to shift attention in a flexible and adaptive manner. Mirsky acknowledges that this behavior is extremely complex and goes beyond what is commonly included in the construct of attention. His views are reflected in his comment that "the boundaries between 'attention' and 'executive function' are rather indistinct" (1991, p.130). Specific areas of the prefrontal cortex represented by this function include the dorsolateral prefrontal cortex, the medial frontal cortex, and the anterior cingulate gyrus.

Finally, encode is the mnemonic component of attention hypothesized by Mirsky. Using data from electrophysiological measures and behavioral observation, he locates mediation of the encode function within the hippocampus and amygdala. This function represents the initial entry point of information into the memory system. Specifically, it affords the individual the ability to hold information briefly for immediate manipulation or action. One might equate this function to that of "working memory", which has been similarly defined. Indeed, the relationship between attention and the ability to encode information into short term memory has been extensively explored. As one might expect, it has proven difficult to tease out differences between these two highly interrelated functions (Cohen, 1993).
**Summary and Hypotheses**

To promote an understanding of attentional functioning in children, Mirsky's taxonomy of attention was explored in children with MM. This inquiry represents the first theory-driven exploration of attention in this population. In addition to utilizing the neuropsychological tasks outlined by Mirsky to explore attentional functioning, data were collected regarding the behavioral manifestation of attention in these children. Academic achievement was also assessed for each child. Finally, a sibling control group was utilized to allow for more accurate analyses of these data.

Specific hypotheses include:

1) Children with MM will score lower on neuropsychological facets of attention as compared to a group of siblings.

2) Attentional facets will be related to the presence of medical complications associated with MM even after controlling for sociodemographic variables.

3) Performance on facets of attention will predict behavioral manifestations of attention as well as academic achievement.
CHAPTER 2

METHOD

Participant Recruitment

All participants were recruited for a broader study of neuropsychological functioning in MM from the MM clinic roster of a large teaching hospital in a midwestern metropolitan area. A recruitment letter was sent to each family whose child with MM was in the designated age range of 8-0 through 15-11 years of age. Letters were followed by a telephone call from the research associate who further explained the study, answered questions, and arranged for an assessment appointment if the family was willing to participate. Families were paid $25 for their participation. They also received a summary report of the assessment findings free of charge. Recruitment of participants continued throughout the course of the 2 year study.

One year into the study, families that had participated initially were again contacted by letter. This letter requested the participation of a biological sibling of the MM child who was also in the designated age range (8-15 years). Siblings were recruited to serve as a control group. All siblings were free of known neurological impairments. To the extent possible, same-sex siblings were included. Once again, a telephone contact was made in order to answer questions and schedule assessment appointments. Families were paid $50 for the sibling assessment, and also received a summary report of assessment findings free of charge.
Demographic data collected on each subject included: age, sex, ethnic group, parental education and occupation, grade in school, type of school placement, a history of grade repetition, and dominant hand preference.

Sample Characteristics

Sixty-four children with MM, 38 males and 26 females participated in the study. They represented 67% of all eligible children. Analyses of subject demographics indicated that participants in the study did not differ from non-participants in age, gender, ethnicity, shunt placement, classroom placement, or parental education (see Table 1).
<table>
<thead>
<tr>
<th></th>
<th>MM Group</th>
<th>Sibling Group</th>
<th>Non-Participant MM Group</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Total N</strong></td>
<td>64</td>
<td>27</td>
<td>48</td>
</tr>
<tr>
<td><strong>Age (SD)</strong></td>
<td>10.95 (2.44)</td>
<td>12.26 (2.05)</td>
<td>12.03 (2.06)</td>
</tr>
<tr>
<td><strong>% Males</strong></td>
<td>59%</td>
<td>56%</td>
<td>42%</td>
</tr>
<tr>
<td><strong>% Caucasian</strong></td>
<td>95%</td>
<td>100%</td>
<td>90%</td>
</tr>
<tr>
<td><strong>% Reg. Classroom</strong></td>
<td>63%</td>
<td>96%</td>
<td>47%</td>
</tr>
<tr>
<td><strong>Parental Education:</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; High School</td>
<td>17%</td>
<td>11%</td>
<td>9%</td>
</tr>
<tr>
<td>H.S. Graduate</td>
<td>52%</td>
<td>54%</td>
<td>67%</td>
</tr>
<tr>
<td>Partial College</td>
<td>9%</td>
<td>6%</td>
<td>9%</td>
</tr>
<tr>
<td>College Graduate</td>
<td>22%</td>
<td>30%</td>
<td>15%</td>
</tr>
</tbody>
</table>

Table 1. Demographics of sample participants and non-participants.
Twenty-seven siblings served as the control group for the study. They represented 77% of all eligible siblings of the total MM sample. The sibling control group included 15 boys and 12 girls, which resulted in a gender match for 74% of the matched sibling pairs. Forty percent (40%) of the children with MM either did not have a sibling or did not have one within the designated age range. No demographic differences were found (e.g., age, gender, classroom placement, parental education, socioeconomic status) between MM children with and without sibling participants. They also did not differ in intellectual functioning on the Verbal Comprehension Index (VC) or the Perceptual Organization Index (PO) of the Wechsler Intelligence Scale for Children (WISC III; Wechsler, 1991), nor on achievement (composite score of the Wechsler Individual Achievement Test (WIAT), Psychological Corporation, 1992). Group comparisons between study participants indicated that while there were no differences between children with MM and sibling controls on gender, race, or SES, the groups had significantly different ages at testing ($T=-2.759, df=69, p<.01$) and different overall intellectual abilities ($T=-9.067, df=69, p<.001$), with the siblings being older and having higher IQ scores.

Procedure

As part of the larger study, a full neuropsychological battery was administered to each child. All tests were administered individually by the research associate of the project. The battery was divided into four sections, the administration of which was randomized to control for order effects. It took approximately 4 1/2 to 5 hours to secure voluntary consent/assent and to complete the assessment process. With two exceptions, all
assessments were conducted over the course of one day, stopping at mid-point for a one hour lunch break. The parent completed several forms regarding the child, including a behavioral questionnaire with ratings of attention.

A comprehensive retrospective review of the medical record for each child with MM was conducted. To check for accuracy, each variable was then reviewed with the director of the MM Clinic at the hospital. This input was particularly valuable since this physician has followed the majority of the children in the study since birth.

The test battery used in this study included measures of Mirsky's four elements of attention, ratings of attentional behavior, and a measure of academic achievement.

Consistent with Mirsky's model, composite scores for the four attentional factors were developed by averaging the age-corrected standard scores across tests within each of the four domains. Specific tasks which comprised the four factors are:

Focus/Execute: Trail Making Test, Parts A and B (Reitan & Wolfson, 1985); Coding subtest, WISC-III (Wechsler, 1991).

Sustain: Continuous Performance Task (Gordon, 1983); scores of Omission (failure to hit a correct target) and Commission (hitting an incorrect target).

Encode: Arithmetic and Digit Span subtests, WISC-III.

Shift: Wisconsin Card Sorting Test (WCST) (Heaton et al., 1993); Percent conceptual responses; Percent perseverative errors.

Measures

Wechsler's Intelligence Scale for Children - Third Edition (Wechsler, 1991). The complete WISC-III was administered including the Digit Span supplemental subtest.
Standardized scores for Verbal, Performance, and Full-Scale IQs were garnered, as well as factor scores for Verbal Comprehension (VC), Perceptual Organization (PO), and Freedom from Distractibility (FD).

The WISC-III has been shown to demonstrate sound psychometric properties. Split-half reliability coefficients for the IQ and factor scores are all .85 or better for children within the age range of this sample. Standard errors of measurement are also adequate, ranging from 3.35 to 6.36 for the IQ and factor scores. Validity has also been demonstrated (Wechsler, 1991).

*Trail Making Test-Parts A and B* (Reitan & Wolfson, 1985). Part A requires the subject to connect numbered circles that are randomly distributed around the page. Numbers are connected in ascending order, from 1-15. Part B consists of circles that contain either numbers or letters. The subject alternates between number and letter, connecting these in ascending order until the final circle is reached. Numbers are from 1-8, letters from A-G. Scores are the time, in seconds, taken to complete each sequence.

This test has been used extensively in neuropsychological assessment. It is described as a test of speeded visual search, attention, cognitive flexibility, and motor function. Part B is associated with the ability to deal concurrently with more than one stimuli, and to switch between mental sets during an activity. Adequate norms for children are available (Spreen & Strauss, 1991).

*Continuous Performance Test: Vigilance Task* (Gordon, 1983). Numbers are displayed on a screen in front of the subject at 1 second intervals for 9 minutes. The stimulus is displayed for 800ms, and there is a 200ms delay between stimuli. The subject
depresses a button each time the target stimuli appears (1 followed by 9). There are a total of 45 targets over the course of the 9 minute period. Scores are the number of omissions and number of commissions.

CPT tasks have long been used as measures of sustained attention (Gordon, 1993). They are one of the few psychological assessment techniques that have reliably distinguished clinical populations of children with ADHD from normal controls (Barkley & Grodzinsky, 1994).

**Wisconsin Card Sorting Test (WCST; Heaton, et al., 1993).** The test is a card sorting task in which 128 cards are sorted according to three principles: color, form, number. Four key cards are placed in front of the subject and he/she is given feedback regarding the accuracy of card placement. Sorting principles are not revealed to the child, and no further directions are given. Scoring includes the percentage of conceptual level responses and the percentage of perseverative errors.

The percent of conceptual response is defined by the ability to maintain at least 3 consecutive correct answers. It serves as an index of insight into the correct sorting strategy. The percent perseverative error score is a measure of the concentration of incorrect perseverative responses in relation to overall test performance. Perseveration is persistent, inflexible responding even in light of evidence that the response set is incorrect or inefficient.

The WCST has been used extensively in neuropsychological evaluations to measure executive function in adults and children. It has been shown to discriminate younger aged children diagnosed with ADHD from normal controls.

Reliability has not been as well researched for this test (Franzen, 1989). However, the most recent manual (Heaton et al., 1993) reports that generalizability coefficients were
modest to good in a sample of normal children and adolescents. It is widely accepted as a sensitive index of brain injury, although localization of focal damage remains controversial (Damasio & Anderson, 1993). Nonetheless, perseverative response or error scores on the WCST are its most sensitive index of frontal lobe dysfunction (Damasio & Anderson, 1993).

**Child Behavioral Checklist - Parent Form (CBCL; Achenbach, 1991).** The CBCL is a 138-item checklist completed by a parent. Normative data are available from 1,300 community non-clinic referred and 2,300 mental health clinic-referred children. Normalized T-scores are available for ages 4-18. Eight specific scale scores are provided, in addition to summarative scores. The score on the Attention Problems scale was utilized in this research.

The CBCL is one of the most widely used standardized rating scales of children’s behavior. The Attention Problem scale demonstrates high inter-rater agreement (r=.79) and good stability across time (r=.75). Validity has been demonstrated via its ability to differentiate clinic referred from non-clinical child samples, as well as its adequate correlations with other measures of child behavior (e.g., Conners Parent Questionnaire) (Achenbach, 1991).

**Wechsler Individual Achievement Test-Screener (WIAT-Screener) (1992).** The WIAT Screener consists of a composite score comprised of three subtests: Reading (word identification), Mathematical Reasoning, and Spelling. Scores are standardized based upon age normative data. In addition, a composite score is achieved by adding the raw scores of the three subtests and determining the standard score from age normed tables.

Split-half reliability coefficients are quite good for the Screener, falling at or above r=.95 for the age of children in this study. Standard error of measurement is also quite low.
Content, construct, and criterion measures of validity are also adequate. Performance on the WIAT by children diagnosed with ADHD has been shown to be average, with the lowest scores found on Basic Reading, Spelling, and Numerical Operations (WIAT Manual, 1992).

**Medical Variables:** Medical variables garnered for each subject were quite comprehensive. To facilitate analysis, a subset of medical variables were chosen for inclusion here based upon the literature and previous analyses of these data (Yeates, Loss, & Enrile, 1994). Variables included in this study were: the presence/absence of shunted hydrocephalus; the number of shunt revisions; high (T12 to L2) or low (L3 and below) lesion level; the presence/absence of partial / complete agenesis of the corpus callosum; a history of CNS infection (ventriculitis); and the presence/absence of oculomotor abnormalities. The overall incidence of each of the medical variables is reported in Table 2.
Table 2. Presence of medical variables in sample of children with MM (N=64).

<table>
<thead>
<tr>
<th>Medical Variable</th>
<th>Total Number</th>
<th>% Total Sample</th>
</tr>
</thead>
<tbody>
<tr>
<td>Shunt</td>
<td>56</td>
<td>87.5%</td>
</tr>
<tr>
<td># Shunt Revisions</td>
<td>45*</td>
<td>80.4%*</td>
</tr>
<tr>
<td>Mean=1.63 SD=1.66</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shunt Infection</td>
<td>6</td>
<td>10.6%</td>
</tr>
<tr>
<td>High Lesion Level</td>
<td>29</td>
<td>45%</td>
</tr>
<tr>
<td>(T12-L2)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Agenesis of Corpus Callosum</td>
<td>15</td>
<td>23%</td>
</tr>
<tr>
<td>Oculomotor Abnormalities</td>
<td>28</td>
<td>44%</td>
</tr>
</tbody>
</table>

* = Total number/percentage of shunted children with at least one shunt revision

---

Data Analysis Plan

Several preparatory steps were taken with the data prior to hypothesis testing. First, inclusion criteria were set for each analysis to ensure the validity of results. Composite scores for the four attention factors were plotted in a stem and leaf format to identify extreme scores which would unreasonably bias the data. Extreme scores were identified as those more than three times beyond the interquartile range (Kirby, 1993). Removal of children with extreme scores resulted in the deletion of two siblings and two children with MM because of extreme scores on "sustain", and one child with MM because of an extreme score on "focus/execute".

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Second, to control for general mental deficiency, participants whose scaled scores were lower than 70 on both the Verbal Comprehension Index (VC) and the Perceptual Organization Index (PO) of the WISC-III were deleted from the analyses of hypotheses 1 and 3, which were concerned with group differences and the prediction of ratings of attentional behavior and academic skills (see Table 3). Based on this criteria, 15 children with MM were deleted. Therefore, the total sample used in the analyses of hypothesis 1 and 3 consisted of 71 children (MM/S=38; MM/NO=8; Siblings, N=25).

Children with MM and low VC and PO scores were not excluded from the analyses of hypothesis 2, which examined the prediction of attention by medical variables. This was done so as not to truncate the range of outcomes and to insure that results would reflect the variability found within a clinical sample of children with MM. However, children with missing data on the composite attention scores were not included in this analysis. Missing data was the result of incomplete testing, and resulted in the exclusion only of children with very low IQ who were also shunted. The total number of subjects used in each analysis was: “focus-execute”, N=60; “sustain”, N=58; encode, N=64; and “shift”, N=57.

Finally, a sub-sample of twenty matched sibling pairs was extracted from the larger data set. All children in this sub-sample met the inclusion criteria outlined above. Children in this group were full biological siblings who were raised in the same home. This sub-sample of 40 children was used in follow-up analyses of hypotheses 1 and 3.
<table>
<thead>
<tr>
<th></th>
<th>MM - Total (N=64) Mean (SD)</th>
<th>MM - Higher IQ (N=46) Mean (SD)</th>
<th>Sibling Controls (N=25) Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Full Scale IQ</td>
<td>73.94 (16.50)</td>
<td>81.59 (11.57)*</td>
<td>108.00 (12.00)</td>
</tr>
<tr>
<td>Verbal Comp.</td>
<td>83.75 (18.41)</td>
<td>91.83 (13.92)*</td>
<td>109.52 (12.85)</td>
</tr>
<tr>
<td>Perceptual Org.</td>
<td>72.97 (16.05)</td>
<td>78.28 (14.45)*</td>
<td>104.84 (12.05)</td>
</tr>
<tr>
<td>WIAT Composite</td>
<td>85.14 (20.43)</td>
<td>93.39 (15.74)*</td>
<td>106.92 (12.48)</td>
</tr>
<tr>
<td>CBCL Attention</td>
<td>60.86 (9.19)</td>
<td>60.33 (9.59)*</td>
<td>52.28 (5.20)</td>
</tr>
<tr>
<td>Focus/Execute</td>
<td>21.55 (25.94)</td>
<td>30.29 (13.99)*</td>
<td>47.56 (10.38)</td>
</tr>
<tr>
<td>Sustain</td>
<td>20.12 (47.40)</td>
<td>35.54 (18.98)</td>
<td>50.97 (5.78)</td>
</tr>
<tr>
<td>Encode</td>
<td>39.90 (9.28)</td>
<td>43.77 (7.24)*</td>
<td>52.27 (6.49)</td>
</tr>
<tr>
<td>Shift</td>
<td>39.27 (9.96)</td>
<td>41.15 (9.48)*</td>
<td>55.50 (9.08)</td>
</tr>
</tbody>
</table>

* Subjects with extreme scores were deleted.
* MM children with higher IQ differ significantly from controls (p<.001)

Table 3. Neuropsychological data for total Myelomeningocele group (MM), higher IQ MM sub-group, and sibling controls

Hypothesis 1. To test for group differences on the four Mirsky factors (hypothesis 1), a multivariate analyses of covariance was conducted (MANCOVA). Covariates used in the analysis were socioeconomic status (SES) (Hollingshead 4 Factor
Index; Hollingshead, 1975) and age. Significant multivariate results were followed by univariate tests, and significant univariate findings were subject to Tukey post hoc analyses. To further elucidate these relationships, group differences were explored for a subset of matched sibling pairs (N=20 pairs). Paired t-tests were used to analyze differences in performance on the four elements of attention for this sub-sample of children.

**Hypothesis 2.** To examine the effect of medical complications associated with MM on the composite attention variables, a series of multivariate regression analyses were conducted. Six medical variables from a larger data set were used to test this hypothesis: shunt placement (yes/no); number of shunt revisions; lesion level (coded as "high" - T12 to L2 or "low" - below L2); partial or complete agenesis of the corpus callosum; history of ventriculitis (CNS infection); and the presence of oculomotor abnormalities (e.g., esotropia, strabismus).

A series of hierarchical regression analyses were conducted with the composite attention scores as the dependent variable. The first step in each analysis was to regress the respective attentional score onto socioeconomic status and age. The second step was to include the six medical variables identified above. Therefore, the final analysis tested the predictive value of all six medical variables, after controlling for demographics, as well as the unique contribution of each medical variable over and above that accounted for by all other variables.
Hypothesis 3. The final question was whether the cognitive measures of attention could predict ratings of attentional behavior and academic achievement. To answer this, a series of multivariate hierarchical regression analyses were conducted. Essentially, the same steps were used for each part of the question.

First, since the sample included both children with MM and siblings, the dependent variable (CBCL Attention Problem scale score; WIAT Composite score) was regressed onto group status for the prediction to behavior, and onto group status and behavior for the prediction to achievement. Second, the four attention composite scores were included. Finally, group by element of attention interaction terms were included in the model to determine if predictive relationships differed for the children with MM as compared to the siblings. If interaction terms were not significant, they were removed from the model and main effects were examined.

We also subjected a sub-sample of matched sibling pairs (N=20 pairs) to the above analyses. Hierarchical regression analyses were again used following the same sequence.
CHAPTER 3
RESULTS

Hypothesis #1

The first hypothesis stated that group differences would be found on the four elements of attention proposed by Mirsky. Results indicated that a significant multivariate group difference did exist (Wilks Lambda $\Lambda(4, 63) = 19.23, p < .001$). Subsequent univariate analyses were conducted for each element of attention while controlling for the effect of age and SES. Significant results were obtained for all four elements: “focus/execute” ($F(2, 66) = 9.96, p < .001$), “sustain” ($F(2, 66) = 4.01, p < .05$), “encode” ($F(2, 66) = 11.61, p < .001$) and “shift” ($F(2, 66) = 14.85, p < .001$). As indicated by $F$ values, the magnitude of difference was more pronounced for the elements of “focus/execute”, “encode”, and “shift”, than for “sustain”.

Tukey honestly significant difference (HSD) post hoc tests, as presented in Table 4, indicated that the siblings performed significantly better than the MM/S group on all measures of attention (“focus/execute” $p < .001$; “sustain” $p < .05$; “encode” $p < .001$; “shift” $p < .001$). In contrast, the siblings performed better than the MM/WO group on “encode” ($p < .001$) and “shift” ($p < .01$), yet not on “focus/execute” ($p = .10$) or “sustain” ($p = .17$). There were no differences between the MM/S and MM/WO groups on any attentional component.
<table>
<thead>
<tr>
<th></th>
<th>MM W/Shunt (N=38)* Mean (SD)</th>
<th>MM W/O Shunt (N=8)* Mean (SD)</th>
<th>Sibling Controls (N=25)* Mean (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Verbal Comp.(^1)</td>
<td>91.50 (13.79)</td>
<td>93.38 (15.41)</td>
<td>109.52 (12.85)</td>
</tr>
<tr>
<td>Perceptual Org.(^2)</td>
<td>77.18 (13.68)</td>
<td>83.50 (17.77)</td>
<td>104.84 (12.05)</td>
</tr>
<tr>
<td>Focus/Execute</td>
<td>29.36 (13.95)*</td>
<td>34.69 (14.28)</td>
<td>47.56 (10.38)</td>
</tr>
<tr>
<td>Sustain</td>
<td>35.31 (17.92)*</td>
<td>36.64 (24.83)</td>
<td>50.97 (5.78)</td>
</tr>
<tr>
<td>Encode</td>
<td>44.17 (7.66)*</td>
<td>41.88 (4.67)*</td>
<td>52.27 (6.49)</td>
</tr>
<tr>
<td>Shift</td>
<td>41.20 (10.23)*</td>
<td>40.94 (4.91)*</td>
<td>55.50 (9.08)</td>
</tr>
</tbody>
</table>

* Controlling for general mental deficiency and outlying data.
\(^1\) Verbal Comprehension Index from the WISC-III.
\(^2\) Perceptual Organization Index from the WISC-III.
* p<.001 different from sibling score
* p<.01 different from sibling score
* p<.05 different from sibling score

Table 4. Performance on Measures of Attention for Shunted and Non-Shunted MM Groups and Sibling Controls
To further elucidate group differences, a series of paired-samples T-tests were conducted on matched sibling pairs. Significant differences were found between the groups on all four of Mirsky’s elements of attention. Results are presented in Table 5.

<table>
<thead>
<tr>
<th>Element</th>
<th>Children with MM</th>
<th>Matched Siblings</th>
<th>t</th>
</tr>
</thead>
<tbody>
<tr>
<td>Focus/Execute</td>
<td>30.563 (12.95)</td>
<td>48.13 (11.38)</td>
<td>4.78*</td>
</tr>
<tr>
<td>Sustain</td>
<td>35.07 (21.14)</td>
<td>50.85 (5.87)</td>
<td>3.16b</td>
</tr>
<tr>
<td>Encode</td>
<td>47.00 (7.83)</td>
<td>53.17 (5.82)</td>
<td>2.45*</td>
</tr>
<tr>
<td>Shift</td>
<td>44.40 (9.98)</td>
<td>55.25 (9.51)</td>
<td>3.57b</td>
</tr>
</tbody>
</table>

*p < .05  
*b < .01  
*c < .001

Table 5. Performance on Elements of Attention for Matched Sibling Pairs (N=40)

Hypothesis #2

The results of these analyses indicated that medical variables are of substantial value when predicting the performance of children with MM, particularly for “focus/execute” and “shift”. This was true even after controlling for demographic influences of age and socioeconomic status (see Table 6).
While a significant amount of variance in “focus/execute” was predicted by the medical and demographic variables together, the medical variables in and of themselves contributed significantly (change in $R^2 = 21.6\%$; $F(6,51) = 2.43; \ p < .05$). Examination of the equation indicated that a history of ventriculitis predicted uniquely to “focus/execute” ($p < .05$), while the unique contribution of oculomotor abnormalities approached significance ($p = .07$).

Approximately twenty-five percent (24.5%) of the variance in “shift” was accounted for by the medical and demographic variables together. As before, tests of effects indicated that there was a significant contribution by the medical variables (change in $R^2 = 23.4\%$, $F(6,48) = 2.48$, $p < .05$). Three medical variables contributed unique variance above that accounted for by the other variables: the number of shunt revisions

<table>
<thead>
<tr>
<th></th>
<th>Demographics Alone</th>
<th>Demographics and All Medical Variables</th>
<th>Percent Change in $R^2$</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$R^2$</td>
<td>$F/p$</td>
<td>$R^2$</td>
</tr>
<tr>
<td>FOCUS</td>
<td>.028</td>
<td>.83</td>
<td>.244</td>
</tr>
<tr>
<td>SUSTAIN</td>
<td>.152</td>
<td>4.95</td>
<td>.271</td>
</tr>
<tr>
<td>ENCODE</td>
<td>.028</td>
<td>.87</td>
<td>.189</td>
</tr>
<tr>
<td>SHIFT</td>
<td>.011</td>
<td>.31</td>
<td>.245</td>
</tr>
</tbody>
</table>

Table 6. Predictive values of demographic and medical variables in children with MM.
the presence of oculomotor abnormalities (p < .05); and partial or complete agenesis of the corpus callosum (p < .05). All predicted poorer performance on “shift”.

Although twenty-seven percent (27.1%) of the variance in “sustain” was accounted for by the entire model, the inclusion of the medical variables was not significant (change in $R^2=11.9\%$, $F(6, 49) = 1.32$, $p > .10$). However, the presence of oculomotor abnormalities was suggestive of poorer performance on “sustain” ($p = .056$).

An additional 15.6% of the variance was accounted for in “encode” by inclusion of the medical variables, yet this also failed to reach significance ($F(6, 55) = 1.82$, $p > .10$). The presence of oculomotor abnormalities ($p < .05$) and a high lesion level ($p < .05$) did account for unique variance in “encode”, while partial or complete agenesis of the corpus callosum approached significance ($p = .09$).

In summary, as a group the medical variables contributed significantly to the explanation of scores for “focus/execute” and “shift”. Individually, several of the medical variables also predicted to those two elements of attention, as well as to “encode” and “sustain”. The presence of medical complications, therefore, is indicative of poorer performance on the attention factors.
Hypothesis #3

The effect of the cognitive elements of attention on ratings of attention behavior was addressed first. The initial two steps of the hierarchical regression analysis (group status; composite scores) revealed that approximately 36% of the variance in the CBCL Attention Problems scale was accounted for by these variables. Tests of effects indicated that the four cognitive composite scores contributed significantly to the equation after accounting for group differences (change in $R^2 = 17.8\%$, $F(4, 65) = 4.52; p < .01$) (Table 7).
<table>
<thead>
<tr>
<th></th>
<th>FULL SAMPLE N=71</th>
<th>MATCHED SIBLINGS N=40</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Regressed onto Group Status</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$R^2$</td>
<td>.180</td>
<td>.175</td>
</tr>
<tr>
<td>$F$ (df) $p$</td>
<td>15.13 (1.69) .001</td>
<td>8.08 (1.38) .007</td>
</tr>
<tr>
<td><strong>Regressed onto Group Status and Attention Elements</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$R^2$</td>
<td>.358</td>
<td>.395</td>
</tr>
<tr>
<td>$F$ (df) $p$</td>
<td>7.26 (5.65) .001</td>
<td>4.44 (5.34) .003</td>
</tr>
<tr>
<td>% Change in $R^2$</td>
<td>17.8%</td>
<td>22%</td>
</tr>
<tr>
<td>$F$, $p$</td>
<td>4.52, .003</td>
<td>3.09, .029</td>
</tr>
<tr>
<td><strong>Regressed onto Group Status, Attention Elements, and Interactions</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>$R^2$</td>
<td>.457</td>
<td>.606</td>
</tr>
<tr>
<td>$F$ (df) $p$</td>
<td>5.7 (9.61) .001</td>
<td>5.14 (9.31) .001</td>
</tr>
<tr>
<td>% Change in $R^2$</td>
<td>9.9%</td>
<td>21.1%</td>
</tr>
<tr>
<td>$F$, $p$</td>
<td>2.76, .035</td>
<td>4.03, .01</td>
</tr>
</tbody>
</table>

Table 7: Prediction of ratings of attentional behavior by Mirsky's elements of attention.
A test of interactions between group (MM, sibling) and the four elements of attention on ratings of attentional behavior was significant (Change in $R^2 = 10\%$, $F(4, 61) = 2.77$, $p < .05$). Of particular interest was the discovery that “focus/execute” was correlated with ratings of attentional behavior for the MM group ($p < .01$), but not for siblings. In contrast, “sustain” was correlated with ratings of siblings’ behavior ($p < .05$) but not with ratings of children with MM (Tables 8 and 9).
Table 8. Correlations of elements of attention with ratings of behavior and academic achievement for higher IQ MM group (N=48).

<table>
<thead>
<tr>
<th></th>
<th>Focus/Execute</th>
<th>Sustain</th>
<th>Encode</th>
<th>Shift</th>
<th>CBCL Attention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Focus/Execute</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sustain</td>
<td>.289</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Encode</td>
<td>.276</td>
<td>.185</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shift</td>
<td>.184</td>
<td>.178</td>
<td>.313</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CBCL Attention</td>
<td>-.498</td>
<td>.074</td>
<td>-.295</td>
<td>-.061</td>
<td></td>
</tr>
<tr>
<td>WIAT Composite</td>
<td>-.317</td>
<td>.092</td>
<td>.545</td>
<td>.302</td>
<td>-.317</td>
</tr>
</tbody>
</table>

Table 9. Correlations of elements of attention with ratings of behavior and academic achievement for sibling group (N=25).

<table>
<thead>
<tr>
<th></th>
<th>Focus/Execute</th>
<th>Sustain</th>
<th>Encode</th>
<th>Shift</th>
<th>CBCL Attention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Focus/Execute</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sustain</td>
<td>.512</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Encode</td>
<td>.141</td>
<td>.400</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shift</td>
<td>.183</td>
<td>.280</td>
<td>.552</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CBCL Attention</td>
<td>.068</td>
<td>-.442</td>
<td>-.108</td>
<td>-.109</td>
<td></td>
</tr>
<tr>
<td>WIAT Composite</td>
<td>-.007</td>
<td>.194</td>
<td>.501</td>
<td>.307</td>
<td>-.202</td>
</tr>
</tbody>
</table>
The second part of this hypothesis stated that the four elements of attention would predict academic functioning beyond that accounted for by individual or group differences in attentional behavior. A significant percent of the variance (23.7%) in academic achievement was accounted for by group status and ratings of attentional behavior. Inclusion of the four elements of attention, however, added significantly (change in $R^2 = 20.5\%$, $F(4, 64) = 5.89; p < .001$). Together, these variables accounted for over forty percent of the variance in academic achievement ($R^2 = .442$, $F(6, 64) = 8.47, p < .001$). “Encode” was discovered to contribute unique variance above that accounted for by the remaining variables ($p < .001$). A test of the interaction between group status and the four elements of attention was not significant ($p = .847$), thereby suggesting that the relationship between “encode” and achievement held for both the MM group and sibling controls (Table 10).
<table>
<thead>
<tr>
<th>Regression Model</th>
<th>Full Sample N=71</th>
<th>Matched Siblings N=40</th>
</tr>
</thead>
<tbody>
<tr>
<td>Regressed onto Group Status and Ratings of Behavior</td>
<td></td>
<td></td>
</tr>
<tr>
<td>( R^2 )</td>
<td>.237</td>
<td>.249</td>
</tr>
<tr>
<td>( F (df) ) p</td>
<td>10.58 (2,68) .001</td>
<td>6.13(2,37) .005</td>
</tr>
<tr>
<td>Regressed onto Group Status, Ratings of Behavior and Attention Elements</td>
<td></td>
<td></td>
</tr>
<tr>
<td>( R^2 )</td>
<td>.442</td>
<td>.536</td>
</tr>
<tr>
<td>( F (df) ) p</td>
<td>8.47 (6,64) .001</td>
<td>6.35 (6,33) .001</td>
</tr>
<tr>
<td>% Change in ( R^2 )</td>
<td>20.5%</td>
<td>28.7%</td>
</tr>
<tr>
<td>( F, p )</td>
<td>5.89, .001</td>
<td>5.11, .003</td>
</tr>
<tr>
<td>Regressed onto Group Status, Ratings of Behavior, Attention Elements, and Interactions</td>
<td></td>
<td></td>
</tr>
<tr>
<td>( R^2 )</td>
<td>.455</td>
<td>.561</td>
</tr>
<tr>
<td>( F (df) ) p</td>
<td>5.01 (10,60) .001</td>
<td>3.7 (10,29) .003</td>
</tr>
<tr>
<td>% Change in ( R^2 )</td>
<td>.013%</td>
<td>.025%</td>
</tr>
<tr>
<td>( F, p )</td>
<td>.34, .847</td>
<td>.41, .804</td>
</tr>
</tbody>
</table>

Table 10. Prediction of ratings of academic achievement by attentional behavior and Mirsky's elements of attention.
The same hierarchical regressions were repeated with the sample of matched sibling pairs. In the first analysis the four cognitive measures of attention contributed significantly to the prediction equation ($F(4, 34) = 3.09, p < .05$), adding approximately twenty-one percent (21%) to the amount of variance accounted for in attentional behavior. The inclusion of interaction terms was significant (change in $R^2 = 21.5\%, F(4,30) =4.03, p <.01$) (see Table 7). As before, “focus-execute” was strongly related to the Attention subscale for the children with MM group ($r = -.572$), while “sustain” was strongly related to behavior for their siblings ($r = -.490$) (Tables 11 and 12).
<table>
<thead>
<tr>
<th>Focus/Execute</th>
<th>Sustain</th>
<th>Encode</th>
<th>Shift</th>
<th>CBCL Attention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Focus/Execute</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sustain</td>
<td>.194</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Encode</td>
<td>.336</td>
<td>.087</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shift</td>
<td>.087</td>
<td>.332</td>
<td>.255</td>
<td></td>
</tr>
<tr>
<td>CBCL Attention</td>
<td>-.572</td>
<td>.336</td>
<td>-.276</td>
<td>.078</td>
</tr>
<tr>
<td>WIAT Composite</td>
<td>.241</td>
<td>-.009</td>
<td>.637</td>
<td>.419</td>
</tr>
</tbody>
</table>

Table 11. Correlations of elements of attention with ratings of behavior and academic achievement for MM in matched sibling pairs (N=20).

<table>
<thead>
<tr>
<th>Focus/Execute</th>
<th>Sustain</th>
<th>Encode</th>
<th>Shift</th>
<th>CBCL Attention</th>
</tr>
</thead>
<tbody>
<tr>
<td>Focus/Execute</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sustain</td>
<td>.629</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Encode</td>
<td>.216</td>
<td>.346</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Shift</td>
<td>-.106</td>
<td>.193</td>
<td>.536</td>
<td></td>
</tr>
<tr>
<td>CBCL Attention</td>
<td>.045</td>
<td>-.490</td>
<td>-.220</td>
<td>-.106</td>
</tr>
<tr>
<td>WIAT Composite</td>
<td>-.075</td>
<td>.184</td>
<td>.529</td>
<td>.321</td>
</tr>
</tbody>
</table>

Table 12. Correlations of elements of attention with ratings of behavior and academic achievement for siblings in matched sibling pairs (N=20).
The ability of the four cognitive elements to predict academic achievement in the matched sibling pairs beyond that accounted for by group and individual behavior differences was substantial (change in $R^2 = 28.7\%$, $F(4, 33) = 5.11, p < .05$) (see Table 10). No element of attention was found to predict to achievement beyond that of the other variables, although “encode” approached significance ($p = .057$). As with the full sample, no differences in prediction were found between groups ($F(4,29) = 0.41, p = .804$).
CHAPTER 4
DISCUSSION

The purpose of this study was to explore the attentional abilities of a well defined child neurological population within a theoretical framework. The efficacy of Mirsky's taxonomy of attention was demonstrated. Children with MM performed more poorly than their siblings on each element of attention. In general, performance on “focus/execute”, “encode” and “shift” was more compromised than performance on “sustain”. A significant relationship was found between medical variables and attention for the MM group, with specific variables correlating more highly with some elements than others. Finally, the four elements of attention predicted to ratings of attentional behavior and to academic achievement. For ratings of attentional behavior moreover, a different predictive pattern was demonstrated for children with MM as compared to a group of their siblings.

Performance Differences on Misky's Elements of Attention

The first question posed in this project was whether or not Mirsky's taxonomy of attention would detect differences between children with MM and a group of their siblings. Indeed, children with MM and shunted hydrocephalus (MM/S) performed more poorly than siblings on each element of attention, even after controlling for general mental deficiency in the MM/S group. A greater magnitude of difference was found between MM/S and siblings on the elements of “focus/execute”, “encode”, and “shift”, while a
lesser magnitude was demonstrated on “sustain”. For children with MM who were contrasted with full biological siblings, performance was significantly more impaired on each element of attention.

It was interesting to note that children with MM who were not shunted (MM/WO) differed significantly from normal siblings only on the elements of “encode” and “shift”. Their scores on “focus/execute” and “sustain” fell between those of MM/S and siblings, and were not significantly different from either group. Although the neuropsychological profile of children with MM is often discussed in terms of the effects of hydrocephalus (Yeates, Enrile, & Loss, in press), these data indicate that factors other than hydrocephalus effect performance on measures of attention. Indeed, the results of hypothesis 2 in this study indicate that oculomotor abnormalities, a history of ventriculitis, and agenesis of the corpus callosum are predictive of poorer attention.

It is also interesting to note that some of the differences found for MM/WO and siblings in this study occurred on tasks that do not have a timed motor demand. This stands in contrast to recent findings in a study of the effects of hydrocephalus of varying etiology on executive function in children. Fletcher, et al., (1996) argue that differential performance on measures of attention in their sample were primarily due to deficits in response speed and motor control. Participants were not restricted exclusively to those with MM, however, and the research was not conducted according to Mirsky’s theory.

It appears therefore, that Mirsky’s taxonomy of attention has provided a more sophisticated way of examining attention in children with MM. Indeed, the robust nature of the model was illustrated by delineating poorer performance on specific elements of attention (“encode” and “shift”) even for children without shunted hydrocephalus, and between biological siblings raised in the same home.
Medical Variables

A recent chapter by Mirsky (1996) reviewed the attentional profiles of adults and children with known neurobehavioral disorders. It was demonstrated that each type of disorder produced a unique profile of attentional deficits. Furthermore, correspondence was found between the known neurological effects of these disorders and the neuroanatomical substrate hypothesized by Mirsky to underlie the facets of attention.

This study evaluated whether medical variables, empirically selected from a larger data base (Yeates, Loss, & Enrile, 1994), could predict performance on Mirsky's elements of attention in children with MM. Results indicated that indeed they could. In this sample, the six medical variables accounted for between 16% and 27% of the variance in each of Mirsky's elements of attention, significantly predicting to "focus/execute" and "shift".

As in Mirsky's review, specific medical variables differentially predicted to the elements of attention. Since the configuration of neurological insults suffered by these children is quite complex, it is challenging to identify the neuroanatomical underpinnings of their attention deficits. Nonetheless, a speculative discussion of how the medical variables might correspond to neurological markers is offered.

Focus/Execute

A history of shunt infection predicted uniquely to performance on "focus/execute". The effects of CNS infection are determined by many factors, and include things such as SES and other demographic variables, age at time of insult, and severity of infection. Although ventriculitis used to be one of the leading causes of death in MM (Lawrence & Tew, 1971), prompt medical response and improved
pharmacological and surgical techniques have significantly diminished its occurrence. Nonetheless, ventriculitus is associated with high morbidity in some studies, and it has been argued that children with a history of ventriculitis should be excluded from analyses, since the effects of ventriculitis are so debilitating that they serve to mask those of any other medical variable (Wills, 1993).

Anatomically, Mirsky (1996) has linked "focus/execute" to the inferior parietal and superior temporal cortices, as well as to the corpus striatum. All three of these structures have complex integrative functions: the inferior parietal area serves to coordinate motor functions; the superior temporal sulcus integrates sensory information; and the corpus striatum receives input from all parts of the cerebral cortex. The integrative role of these brain structures therefore, may indeed make "focus/execute" uniquely vulnerable to the diffuse effects of CNS infection.

Shift

Three medical variables were of unique value in predicting performance on "shift": agenesis of the corpus callosum; oculomotor abnormalities; and total number of shunt revisions. One possible explanation to account for the effect of these variables is to view them as indices of white matter brain damage. White matter disorders are known to diminish the general efficiency of the brain. For these children in particular, the white matter issues may be related to the effects of hydrocephalus.

The relationship between hydrocephalus and damage to white matter has been well established (Del Rigo, 1993). Indeed, hydrocephalus is known to cause stretching, hypoplasia (thinning), and destruction of corpus callosal fibers (Fletcher et al., 1992). For example, hypoplasia effects the posterior body and splenium of the CC; white matter tracts
associated with sensorimotor integration, higher-order cortical functions, and visual areas of the occipital lobes (Fletcher et al., 1992). Further, symptoms of hydrocephalus are the most likely precipitants to a shunt revision (Wills, 1993).

Incomplete or total absence of the CC can also result from disorders of neuroembryogenesis. Associated neuronal migration problems include polymicrogyria, heterotopias, and disrupted myelination (Gilbert, Jones, Rorke, Chernoff, & James, 1986). It is possible therefore, that the predictive value of agenesis of the CC is unrelated to the effects of hydrocephalus. Its predictive value in this sample may be its reflection of significant abnormal brain development.

Performance on the Wisconsin Card Sorting Test, a complex problem-solving task used to measure "shift", requires one to flexibly switch between response alternatives in response to changing demands. Poor performance on this task has been associated with children with spina bifida (Fletcher et al., 1996) and with lesions in the prefrontal cortex (Cohen, 1993). The prefrontal cortex is believed to support the "shift" function (Mirsky et al., 1991) since its extensive reciprocal connections with most brain areas affords it the necessary input and control to switch across response alternatives (Cohen, 1993). It seems reasonable to expect that the underlying cause of disturbances to the CC and ocular abilities may also impact the more phylogenetically more advanced prefrontal cortex.

In this group of children, three variables were able to differentially predict to the "shift" element of attention. All of them are indices of severe neuropathology, either as the result of neuroembryologic dysmorphology, or the effects of significant hydrocephalus. The importance of differentiating the underlying neurological issues has been emphasized recently in a study of attention and executive functions in children with early onset hydrocephalus (Fletcher et al., 1996). Indeed, children with spina bifida who were shunted
and those with evidence of arrested hydrocephalus which was not shunted demonstrated less cognitive flexibility and a greater number of errors than children with spina bifida who had no evidence of hydrocephalus on MRI scans.

**Encode**

Finally, although the contribution of the medical variables as a whole did not reach statistical significance, oculomotor abnormalities and lesion level each predicted uniquely to "encode". Both of these features have been associated with more pronounced dysfunction in the MM population. For example, higher lesion levels are associated with a greater number of physical and cognitive complications (Wills, 1993), and oculomotor abnormalities have been cited as an index of overall neurobehavioral impairment (Zeiner et al., 1985). It is possible therefore, that "encode" is uniquely predicted by these variables simply as an index of the severity of cognitive impairment.

Alternatively, Mirsky linked "encode" directly to the amygdala and the hippocampus, both deep cortical structures which receive input from multiple areas of the brain, including reciprocal connections with brain stem structures. Their location may make their afferent and efferent fibers particularly vulnerable to the Arnold-Chiarri II malformation; which involves caudal displacement and distortion of the brain stem. The neurodevelopmental alterations in these tracts may negatively impact the auditory working memory associated with "encode".

The above explanation is weakened by the fact that children with MM do not typically demonstrate deficits in auditory tasks (Wills, 1993; Yeates, Enrile, & Loss, in press). A different explanation is generated when one explores the demands of the tasks used to measure "encode". While the WISC-III Digit Span and Arithmetic subtests are
believed to reflect auditory working memory, both also require mental manipulation of numbers. Impairment in this ability has been linked to parietal areas (Luria, 1973). It could be that children with MM do poorly on "encode" not because they have poor auditory working memory, but rather because their documented spatial reasoning deficits preclude successful execution of simple math operations.

Sociodemographic Variables

Consistent with previous literature is the finding that sociodemographic variables of socioeconomic status (SES) and age also accounted for unique variance in attention for this sample. In particular, age in years influenced performance on "sustain", while SES influenced performance on "encode". These variables were included in this study since demographic variables have routinely been one of the best predictor of neuropsychological outcome across populations (Taylor, Schatschneider, & Rich, 1992). However, with few exceptions (Fletcher et al., 1996), they have not routinely been reported and considered in neuropsychological studies of children. The importance of considering the impact of economic and familial variables has been argued elsewhere (Yeates, Enrile, & Loss, in press), and is further demonstrated by these results.

Prediction to Behavior and Academic Achievement

The final line of inquiry dealt with the ability of Mirsky's model to predict to ratings of attentional behavior and academic achievement in children. For this sample of children, over 30% of the variance in behavioral ratings of attention and approximately 20% of the variance in academic achievement was accounted for by the four facets of attention in Mirsky's model. These differences were robust even after controlling for
general mental deficiency and within matched sibling pairs. Once again, specific elements of attention were found to predict differently to behavior based upon the neurological status of the child.

**Behavioral Ratings of Attention**

The high prevalence of ADHD and the heterogeneity of its presentation has prompted questions regarding the unitary nature of this disorder. As discussed previously, research has become more sophisticated in its attempt to address these issues by trying to determine the validity of ADHD subtypes. Of central concern to this debate is whether ADHD is a disorder of attention or of behavior. Furthermore, the most valid means of evaluating the diagnostic criteria subsumed under the ADHD rubric have been questioned.

While the diagnostic criteria of ADHD are behavioral in nature, research into the existence of ADHD subtypes has relied almost exclusively on the differential performance on neurocognitive measures of attention. This has prompted investigators to question the validity of this approach (Fletcher, Shaywitz and Shaywitz, 1994). For example, Barkley has argued that the defining characteristic of ADHD is not disordered attention per se, but rather a deficit in rule governed behavior (1994). As such, he contends that the ecological validity of the diagnosis will improve only when measures consistently tap behavioral issues, such as patterns of contingent responding.

The data from this study were analyzed to address this provocative question. Analyses were designed to see if Mirsky's neurocognitive model of attention could predict to behavioral ratings of attention for children, and if neurological status would affect its ability to do so.
Positive results were found. The performance on cognitive measures associated with Mirsky's model did predict to behavioral ratings of attention. Of particular interest however, was the finding that "focus/execute" was a better predictor of behavioral ratings for the children with MM while "sustain" was a better predictor of behavioral ratings for siblings. Therefore, not only did the performance on neurocognitive measures of attention have general predictive capability, specific elements of attention correlated differentially with ratings of attentional behavior based upon the neurological status of the child.

These results can be further discussed within the context of the neuropsychological literature on ADHD subtyping. In this sample, the pattern of attentional performance in children with MM is similar to that identified for children with ADD/WO. Performance of children with ADD/WO is characterized by slowed cognitive and motor output, and a diminished ability to automatize information, true of children with MM as well (Yeates, Enrile, & Loss, in press). The deficit found in ADD/WO has been characterized as one relating to selective attention. "Focus/execute" taps the ability to select relevant stimuli from a broad array and to rapidly initiate action. Poor performance on measures of this component in children with MM are consistent therefore, with the findings that ADD/WO reflects a defect in selective attention. Neuroanatomical findings are also consistent for these two groups, as "focus/execute" has been associated with the inferior parietal area, and deficits in selective attention are linked to the right posterior parietal areas (Mirsky, 1991; Posner & Peterson, 1990). Indeed, it has recently been argued (Fletcher et al., 1996) that the deficits seen in children with hydrocephalus and spina bifida on performance of problem-solving tasks is related to underarousal. This deficiency in arousal would implicate a relative inefficiency in the subcortical, right posterior areas of the brain.
In contrast, ADD/H children are characterized by greater impulsivity, impaired rule-governed behavior, and excessive levels of physical activity (Barkley, 1994). Deficits in sustained attention have been associated with the ADD/H group as a result of their poor performance on CPT tasks. While sustained attention has been linked to primitive areas of the brain (Mirsky, 1991; Posner, 1990), evidence to date suggests that children with ADD/H have differences in frontal brain structures and functions (Goodyear & Hynd, 1992). These discrepant findings may best be understood in the context of Barkley's views. Barkley (1994) believes that poorer performance on the CPT by children with ADHD may actually reflect deficits in rule-governed behavior and rapid habituation to stimuli. Evidence in support of his position is the fact that children with ADHD can attend to stimuli over extended periods of time if they are highly rewarding (e.g., computer games). Within this context, subtle differences in the frontal lobes of children with ADD/H is better explained.

It is interesting that “sustain” was a better predictor of ratings of attentional behavior in siblings, which is consistent with the ADD/H literature. Perhaps this is because the siblings were free of overt neurological impairments, which is true of the general population of ADD/H children as well. It is possible that children with MM may also have frontal differences, yet the phylogenetically more primitive brain areas effected in MM may over-shadow their importance as predictors of problematic attentional behavior.
Academic Achievement

A final test of the utility of Mirsky's model was to test its validity in predicting academic achievement. Specifically, we wanted to see if neurocognitive assessment of attention would offer any predictive value beyond that accounted for by ratings of attentional behavior.

Once again, positive results were found. While ratings of behavior were of value in predicting academic achievement, inclusion of neurocognitive measures of attention increased the amount of variance accounted for in achievement by approximately 20%. "Encode" added uniquely to the predictive equation, consistently so for children with MM and for siblings.

Performance on "encode" is related to the ability to hold information in auditory working memory until some operation can be performed on it. If one thinks about the activity in a typical classroom, the central importance of this ability to academic success is easily seen. Much of the information in school is presented to children in verbal form: lectures, discussions, and directions regarding assignments and classroom routine, for example. An inability to attend to this information long enough to process it would seriously impede the academic performance of any child.

The "encode" score is a composite of performance on the WISC-III Digit Span and Arithmetic subtests. The achievement score is a composite of performance on the Reading, Spelling and Mathematical Reasoning subtests of the WIAT-Screener. Although both of these measure mathematical ability, the correlation between scores may be due in part to the fact that all of these tasks require symbolic manipulation.
Limitations and Future Directions

The results of this study are satisfying, yet improvements can be made on the study design in the future. First, we did not explore the existence and importance of Mirsky’s fifth element of attention, “stability”. Indeed, this study was initially conceptualized and implemented prior to Mirsky’s advance of this idea (Mirsky, 1996). However, it would be interesting to explore these data in reference to this component of attention. Mirsky suggests that performance on the CPT is an adequate measure of this construct, specifically scores of the variability of reaction time, and errors to targets on the AX version. While the AX version of the CPT was included in our test battery, reaction time data are not available. It is possible that the “stability” component of attention will help to further elucidate subtypes of ADHD.

Although a large percentage of children from this hospital’s MM clinic participated, the total number of participants precluded certain types of data analyses. In particular, numbers were insufficient to replicate Mirsky’s factor structure in a robust manner. Additional neuropsychological data gathered from multiple sites, or on-going collection from this site, would bolster the data set and allow for more sophisticated analyses. As stated, Mirsky’s factor structure could be examined, as well as path analyses of the relation between the elements of attention, ratings of attentional behavior, and academic achievement. Finally, a larger sample of children with MM who were not shunted would prove beneficial in teasing out which medical variables best predict performance on measures of attention in this heterogeneous population.

We were fortunate to have access to the physician who ran the MM clinic and knew the majority of children in this study since birth. This allowed us to place greater confidence in our retrospective review of their medical records. Nonetheless, our medical
variables were gross indices of clinically relevant abnormalities or events. In the future, it would be beneficial to have more refined indexes of neurological and neuroanatomical factors in children with MM. For example, access to MRI data could be used to elucidate the extent of Arnold Chiari II malformation and corpus callosal differences. In turn, this would afford greater sophistication in linking performance on measures of attention to underlying neuroanatomical substrates. Although MRI data is expensive to collect, the benefits of this technology in understanding the neuropsychological performance of children have been demonstrated (Fletcher et al., 1996).

Summary

This study presents a theory-driven, systematic exploration of attention in a well defined child neuropsychological population. These results support the robust nature of Mirsky's model in the exploration of attention in children with a known neurological defect. Group differences were demonstrated on performance of each element of attention, with the children with MM and shunted hydrocephalus demonstrating poorer performance than non-shunted children with MM and non-affected siblings. Select medical variables were demonstrated to predict to attentional performance, although the complex nature of MM makes it difficult to tease out the unique contribution of specific variables to distinct elements of attention. Finally, the model was successful in predicting behavioral ratings of attention and academic achievement.

The benefit of viewing attention as a complex system of functions was also supported. Ratings of attentional behavior were not consistently predicted by the same element of attention, in that patterns of performance differed for children with MM and those without. This is particularly significant when one recognizes that behavioral...
measures of attention are most often used to substantiate a diagnosis of ADHD. Indeed, these results suggest that while overt behaviors may be rated similarly, the underlying element of attention which affects the behavior can be different.

The long held clinical impression that attention is impaired in children with MM was empirically documented. The results of this study join a growing body of knowledge regarding attentional functioning in children with known neurological insult. It is hoped that this will promote overall understanding of attention in this age group.
REFERENCES


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