BEHAVIOR PATTERNS AMONG CHILDREN WITH A HISTORY OF METOPIC SYNOSTOSIS

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Metopic synostosis is a condition in which the metopic suture of the human cranium fuses prematurely and may be related to poor behavioral inhibition leading to behaviors commonly associated with Attention-Deficit Hyperactivity Disorder (ADHD). The purpose of this project was to examine the behavior patterns among children with a history of metopic synostosis. It was hypothesized that children with a history of metopic synostosis would exhibit many of the same behavioral patterns associated with ADHD. It was also hypothesized that children with a history of simple synostosis not involving the metopic suture would not evidence this type of behavioral pattern. In order to test these hypotheses, the behavior of three groups of children was compared including (1) children who had a history of metopic synostosis (M= 7.63 years, SD = 1.92 years), (2) children who had a history of simple craniosynostosis not involving the metopic suture (M= 7.54 years, SD = 1.88 years), and (3) a group of children diagnosed with ADHD (M=7.78 year, SD = 1.87 years). It was found using the Home and School versions of the Attention Deficit Disorders Evaluation Scale (ADDES) that children with a history of metopic synostosis demonstrate significantly more behavioral disturbances than children with a history of simple craniosynostosis not involving the metopic suture. Using the BASC Teacher Rating Form it was found that children with a history of metopic synostosis have a behavior pattern similar to children diagnosed with ADHD and a dissimilar behavior pattern compared to children who have a history of craniosynostosis not involving the metopic suture. Using the BASC Parent Rating Form it was found that children with a history of metopic synostosis have a behavior pattern dissimilar to children diagnosed with ADHD and a dissimilar behavior pattern compared to children who have a history of craniosynostosis not involving the metopic suture.

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LITERATURE REVIEW

Craniosynostosis is the premature fusion of one or more of the six main cranial sutures that occurs in approximately 0.4/1,000 births (Hunter & Rudd, 1976). These sutures include the sagittal suture, metopic suture, two bilateral lamdoid sutures and two bilateral coronol sutures. The presence of these sutures allows for the enlargement of the skull vault that expands in order to accommodate a brain weight that doubles during the first year of life. Craniosynostosis often results in a cosmetic deformity of the skull because the premature fusion of one or more of the sutures leads to reduced growth of the cranium perpendicular to the fusion. In order to correct this problem, surgery is frequently performed during the first eighteen months of life.

Craniosynostosis has typically been classified using a description of the suture involved or the resulting head shape. Scaphocephaly or sagittal synostosis is one of the most common forms of craniosynostosis and is characterized by an elongated head from front to back and a palpable ridge along the fused suture. Plagiocephaly involves one of the coronal sutures or lambdoid sutures and is characterized by an asymmetric flattening of the forehead. Trigonocephaly or metopic craniosynostosis is a relatively rare form of craniosynostosis comprising only approximately 3-4% of all cases of craniosynostosis (Hunter & Rudd, 1976). A triangular-shaped forehead and orbital hypotelorism characterize this form of craniosynostosis. Lastly, a spherical-shaped head and a loss of the supraorbital contour on both sides of the head characterize brachycephaly or bicoronal synostosis.

Craniosynostosis can be further classified according to its complexity and etiology. The term “simple synostosis” or “isolated synostosis” is used when a single suture is involved while the term “complex synostosis” is used when more than one suture is involved. The term “syndromal synostosis” is used to refer to a condition in which there are primary defects in morphogenesis and more than one structural abnormality is present. Finally, “secondary synostosis” is used to describe a condition that is the result of one of many disorders such as hyperthyroidism, hydrocephalus etc. (Cohen, 1986).

The underlying etiology of craniosynostosis is very poorly understood. Traditionally, there have been three general theories concerning the pathogenesis of this disorder. Virchow (1981, cited in Cohen, 1986) postulated that the affected suture is the site of the abnormality and deformities of the cranial base occur secondary to this abnormality. In contrast, Moss (1959, cited in Cohen, 1986) suggested that an abnormality in the cranial base is the primary site of dysfunction and deformity of the cranial suture occurs secondary to this abnormality. Third, Park and Powers (1920, cited in Cohen, 1986) believed that the primary defect involves the mesenchymal blastema, which are cells that give rise to connective tissue, of both the cranial base and cranial sutures.

More recently, Martinez-Lage, Poza, Lluch (1996) offered an alternative theory regarding the pathogenesis of craniosynostosis. These researchers reviewed the records of 40 patients with craniosynostosis seen at their hospital over a period of 22 years. They found that three of these 40 patients had associated neural tubes defects. This is the defective closure of the neural tube during early embryogenesis. The authors suggest that the neural tube defects may have acted as a buffer for normal brain pulsation and blocked the intracranial pressure that is believed to stimulate cranial growth. These authors also suggest that craniosynostosis may result from a lack of normal brain pulsation stemming from a lack of cerebral expansion.

Marsh, Koby, and Lee (1993) (cited in Sidoti, Marsh, Marty-Grames and Noetzel, 1996) also presented evidence that children with a history of metopic synostosis may suffer from a lack of prefrontal cortex expansion. These authors conducted a study utilizing three-dimensional brain surface topographic imaging from MRI scans with three children who had a history of metopic synostosis. These researchers found that all three children evidenced similar brain anomalies including abnormally small frontal lobes, excessive subarachnoid cerebrospinal fluid in the anterior fossa and widened pre-central sulci compared to age-matched controls.

Sidoti, Marsh, Marty-Grames and Noetzel (1996) believe that the Marsh, Koby, and Lee (1993) findings are consistent with primary hypoplasia of the brain or neural tissues in the frontal lobes rather than secondary effects of anterior fossa constraint. These authors (Sidoti, Marsh, Marty-Grames and Noetzel, 1996) are quick to note that the small sample size of the Marsh, Koby, and Lee (1993) study precludes the possibility of drawing any firm conclusions. Nevertheless, the findings of Marsh, Koby, and Lee (1993) are especially interesting given the observations and suggestions made by Martinez-Lage, Poza, Lluch (1996). Again, these researchers (Martinez-Lage, et al., 1996) suggested that craniosynostosis may result from of a lack of cerebral expansion of the region underlying the fused suture. This suggestion is also consistent with the view of Sidoti, et al. (1996) that the cerebral anomalies observed in children with metopic synostosis are consistent with primary hypoplasia rather than the secondary effects of anterior fossa constraint.

There is also evidence that suggests craniosynostosis leads to secondary abnormalities of underlying cerebral cortex. Sen et al. (1995) examined the cerebral blood flow distributions in children with a history of simple craniosynostosis. These researchers found regional hypovascularity in the underlying cerebral cortex corresponding to the fused sutures. David, Wilson, Watson, and Argenta (1996) demonstrated that preoperative craniosynostosis is frequently associated with decreased blood flow to cerebral areas directly underlying fused sutures. These authors noted that the cerebral blood flow normalized following surgery and suggested that early surgical intervention is necessary in order to prevent any potential central nervous system compromise that may arise from abnormal blood flow. Rifkenson et al. (1995) also demonstrated that children with craniosynostosis exhibit higher incidences of increased cranial pressure adjacent to the areas of cranial synostosis.

Given this evidence, there are two plausible reasons that children with a history of craniosynostosis may suffer from dysfunction of the cerebral cortex underlying the prematurely fused suture. First, there may be secondary effects from the premature fusion of the cranial suture. Most notably, abnormal cerebral blood flow and hypovascularity (David, Wilson, Watson, and Argenta, 1996; Sen et al., 1995) have been associated with the cerebral cortex underlying prematurely fused sutures. Hypovascularity may lead to a lack of cerebral development in the underlying cerebral cortex. However, it also appears that craniosynostosis itself may be a secondary effect stemming from the lack of cerebral expansion and hypoplasia of the underlying cerebral cortex (Martinez-Lage, Poza, Lluch, 1996; Sidoti, Marsh, Marty-Grames and Noetzel, 1996). Given these two possibilities, it is reasonable to suggest that children who have a history of metopic synostosis suffer from prefrontal cortex dysfunction. It is also reasonable to suggest that there are significant neuropsychological and neuro-behavioral consequences stemming from this prefrontal cortex dysfunction. Consequently, a brief discussion concerning the prefrontal cortex and its associated neuropsychological and neuro-behavioral functions is warranted.

Prefrontal Cortex

The prefrontal cortex is divided into three primary areas that are designed to mediate cognitive, emotional, and motivational processes. These three areas include the dorsolateral prefrontal cortex, the inferior prefrontal cortex and the medial frontal cortex. Each of these prefrontal areas receives and relays information to specific subregions of the striatum, globus pallidus, and thalamus to create prefrontal-subcortical circuits.

The dorsolateral prefrontal circuit originates in the association cortex and has connections with cortical and subcortical regions. These connections include bi-directional communication with the posterior temporal, parietal and occipital association areas. Posterior cortical input reaches the frontal lobe through the inferior longitudinal fascicles of the occipital lobe and the superior longitudinal fasciculus of the parietal lobe. The dorsolateral prefrontal subcortical circuit also serves as a duct for information that is being sent from the dorsolateral portion of the caudate nucleus to the dorsolateral prefrontal cortex (Nieuwenhuys, Voogd, and van Huijzen, 1988).

The inferior prefrontal circuit originates in the orbital frontal cortex and projects information to the ventral portion of the caudate nucleus. This portion of the caudate nucleus receives information primarily from the amygdala as well as information from the substantia nigra, thalamic nuclei and the temporal lobe. The inferior prefrontal cortex also receives information from the dorsolateral prefrontal regions (Nieuwenhuys, Voogd, and van Huijzen, 1988).

The medial frontal subcortical circuit originates in the anterior cingulate cortex and projects to the nucleus accumbens. The anterior cingulate has association connections with the dorsolateral prefrontal cortex. The nucleus accumbens receives input from the amygdala, ventral tegmental area, substantia nigra, and the mediodorsal nucleus of the thalamus (Nieuwenhuys, Voogd, and van Huijzen, 1988).

The overall function of the prefrontal cortex is to mediate behavioral responses in the presence of complex environmental contingencies and in the absence of direct perceptual cues and immediate reinforcement. That is, the frontal lobes mediate contingencies in the absence of guiding environmental cues so that an appropriate behavior can occur. In order for this to occur, each of the prefrontal subcortical units just described perform specific cognitive activities that enables appropriate behaviors to occur in the absence of environmental cues.

The dorsolateral prefrontal subcortical circuit mediates executive behavior and the disruption of this circuit produces executive impairments. These executive impairments are characterized best by an inability to bridge past environmental contingencies to present circumstances. This ability has also been referred to as cross-temporal bridging (Fuster, 1989) and deficits with it can be observed during delayed-response tasks.

Delayed response tasks refer to a class of time-based and response based laboratory procedures. An example of a basic delayed-response procedure using primates involves placing a piece of food under one of two stimuli that are identical except for their spatial location. This is done out of reach, but within full view of the experimental subject. Once the food has been placed under the stimulus, a screen is lowered so that the stimulus (one containing food; one empty) is out of view from the subject. The screen conceals the stimuli for a variable amount of time (usually between 0 and 60 seconds).

Once the screen is removed the experimental subject is given the opportunity to remove one of the stimuli. If the subject removes the stimulus that was covering the food the subject is rewarded with the food. If the subject removes the stimulus that is not covering the food the subject is not reinforced. This task is time based in that the subject must delay a choice until the opportunity to respond is made available. Monkeys that have received lesions to the dorsolateral prefrontal cortex have been found to have impaired performances on this task (see Oscar-Berman, McNamara, and Freedman, 1991 for review). Other deficits that have been associated with disruption to the dorsolateral prefrontal cortex area include reduced verbal fluency, poor abstraction, and impaired response inhibition.

The inferior prefrontal subcortical circuit mediates socially modulated behavior. Disruption of this circuit results in disinhibited, tactless and impulsive behavior. Disorders of the subcortical structures of the inferior prefrontal subcortical circuit, such as Huntington's disease and postencephalitic Parkinson's disease, are often associated with these types of behaviors. In addition, it has been observed that patients that have suffered injuries to this prefrontal area have difficulty shifting sets on the Wisconsin Card Sorting Task. The medial frontal subcortical circuit mediates motivation. Disruption of this circuit is associated with apathy, reduced interest and motivation and an impaired ability to persist on tasks. In addition, impaired inhibition of responses has been observed on go-no go tasks which are tasks in which a response must be inhibited (Drewe, 1975).

Behavioral Disturbances Associated with Prefrontal Cortex Dysfunction

There has been only one study that has attempted to examine the neuro-behavioral consequences associated with metopic synostosis. Sidoti, Marsh, Marty-Grames and Noetzel (1996) conducted a study designed to examine cognitive and behavioral disturbances in children with a history of isolated metopic synostosis. This study was purely descriptive in nature and included 32 cases of children who had a history of metopic synostosis. Utilizing a parent questionnaire developed by the researchers, it was found that four of these patients were identified as having "attention deficit/hyperactivity disorder." Also, four patients were described as being "mentally retarded" and three were described as having "speech and language delays." Despite the fact that this study was purely descriptive in nature and did not utilize a control group, the authors were impressed by the incidence of behavioral disturbances and cognitive impairments found in this study.

It has been hypothesized that disruption of function of the prefrontal cortex may contribute to behavioral disturbances observed in children. More specifically, Barkley (1997) proposed that many of the behavioral disturbances associated with attention deficit hyperactivity disorder stems from dysfunction of the prefrontal cortex. In support of this contention, several studies have demonstrated that people who have been diagnosed with ADHD exhibit many of the same behavioral disturbances as people who are known to suffer from prefrontal cortex dysfunction.

Poor emotional self-control is frequently observed with patients that have suffered prefrontal lobe injuries. Behaviors often associated with poor emotional self-control include irritability, hyper-reactivity and low frustration tolerance (Rolls et al., 1994). Behaviors such as these are considered to be emotional hallmarks of ADHD and have been documented to occur more frequently in children diagnosed with ADHD.

Pelham and Bender (1982) found that children with ADHD are more negative and emotional in their interactions with others compared to normal controls. In addition, several researchers have found a relationship between emotional control and other symptoms associated with ADHD. Shoda et al. (1990) found a significant relationship between poor inhibition on a resistance to temptation task and parents ratings of emotional control in children and frustration tolerance in adolescents. Eisenberg et al. (1993) also found a significant relationship between negative emotional intensity and teachers' ratings of interference control.

It also has been demonstrated that patients who have sustained injuries to the prefrontal cortex are less able to engage in rule-governed behaviors (Delis et al., 1992; Verin et al., 1993). Similarly, there has been a strong indication that children diagnosed with ADHD are less able to engage in rule-governed behaviors. August (1987) found that children diagnosed with ADHD, when given a strategy to use during a memorization task typically discontinued using the rule after only a few trials. It was also found that these same children were less likely to generate their own strategies compared to normal controls (August, 1987). Douglas and Benezra (1990) obtained similar results. Conte and Regehr (1991) also found that children diagnosed with ADHD were less able to transfer rules initially learned in one situation to a novel situation with similar environmental contingencies.

In addition to these behavioral disturbances, it also has been demonstrated that people diagnosed with ADHD exhibit deficits on neuropsychological tests that are believed to be sensitive to prefrontal cortex dysfunction. Boucagnani & Jones (1989) found that 28 children diagnosed with ADHD made significantly more perseverative errors and perseverative responses on the Wisconsin Card Sorting Task than a group of 28 normal children that were matched in terms of age and gender. Interestingly, Berman et al. (1995) have shown that this test is associated with the activation of the dorsolateral prefrontal cortex and is sensitive to frontal lobe damage. Furthermore, Milner (1995) most recently noted that perseverative responses are very common in people that have suffered prefrontal lobe injuries.

Barkley (1992) reviewed 13 studies that examined the performance of patients with ADHD on the Wisconsin Card Sorting Task and found that eight of the studies demonstrated that ADHD patients performed significantly more poorly than normal controls. Barkley (1992) also noted that older subjects were used in the five studies that did not find significant differences between patients diagnosed with ADHD and normal controls. Consequently, Barkley (1992) suggested the prefrontal cortex dysfunction associated with ADHD is subtle in nature and may become less detectable on tests sensitive to prefrontal cortex dysfunction with maturation.

It has also been found that children with ADHD perform poorly on the Stroop Color-Word Interference Test. Impairments are especially evident on the interference portion of this test. This task requires subjects to process a single characteristic of a stimulus (i.e. the color) while simultaneously ignoring another salient characteristic of the stimulus (i.e. the word). Grodzinsky and Diamond (1992) found that a group of 66 children diagnosed with ADHD evidenced significantly more interference on this task than a group of 64 age and gender matched normal children. Gorenstein, Mammato, and Sandy (1989) obtained similar results. The results of these examinations are especially noteworthy in light of the fact that this test has been associated with the activation of the orbital-prefrontal regions (Bench et. al., 1993; Vendrell et al., 1995).

The Trail Making Test, Part B, is another test that has been assumed to be associated with prefrontal cortex functioning. Several studies have shown that children diagnosed with ADHD perform significantly more poorly on this task than normal controls. Gorenstein, Mammato, and Sandy (1989) found that children diagnosed with ADHD perform this task significantly more slowly than normal controls. Boucagnani and Jones (1989) found similar results. In addition, Chelune, Ferguson, Koon, and Dickey (1986) found that a group of children diagnosed with ADHD performed this task significantly more slowly and made significantly more errors than normal controls.

All of the tests described in this section assess the ability to inhibit motor responses while simultaneously performing another task. It is believed that the prefrontal cortex mediates the ability to perform tasks such as this. Consequently, the evidence provided by these studies lends strong support for the hypothesis that underlying prefrontal cortex dysfunction is responsible for the lack of behavioral inhibition observed in children with ADHD.

Modern neuroimaging techniques have also given strong support for the hypothesis that underlying prefrontal cortex dysfunction is responsible for the behavioral disinhibition observed in children diagnosed with ADHD. Geidd, Castellanos, Xavier, Casey, Kozuch (1994) conducted a study designed to examine structural abnormalities of the corpus callosum in children with ADHD. In this study, magnetic resonance imaging was used to measure the corpus callosum in 18 boys diagnosed with ADHD and 18 age and gender matched normal controls. In addition, the parents and teachers of all subjects completed a standardized behavior rating scale. Results indicated that the anterior regions of the corpus callosum were significantly smaller in children with ADHD. In addition, there was a negative correlation between the size of these areas and behavioral disturbances as measured by the standardized behavior rating scales.

Many of these same researchers (Casey, Castellanos, Xavier, Geidd, and Marsh, 1997) recently examined the relationship between prefrontal cortex function and response inhibition in a group of children diagnosed with ADHD and a group of age-matched controls. All subjects were scanned using magnetic resonance imaging (MRI) during three separate response inhibition tasks. The group of children diagnosed with ADHD performed significantly more poorly on all three inhibition tasks compared to the normal control group. In addition, significant correlations were found between the individual performances on the response inhibition tasks and measures of prefrontal cortex dysfunction in the children diagnosed with ADHD. Correlations were strongest for the right hemisphere compared to the left hemisphere.

Similar results were found by Castellanos et al. (1996). This study examined 57 boys that had been diagnosed with ADHD utilizing morphometric magnetic resonance imaging and compared the results with 55 age-matched normal controls. Results indicated that the boys diagnosed with ADHD evidenced significantly decreased volumes of the prefrontal cortex as well as the caudate nucleus, and the globus pallidus. Again, this trend was especially true for the right hemisphere.

Finally, Hynd et al. (1993) found a relationship between the asymmetry of the caudate nucleus and ADHD. Utilizing MRI these investigators examined the head of the caudate nucleus in 11 subjects diagnosed with ADHD and 11 normal controls. Results indicated that normal subjects evidenced a left-larger-than-right asymmetry of the caudate nucleus head while subjects diagnosed with ADHD evidenced a reversal of this pattern. Investigators noted that the reversal of asymmetry in the subjects diagnosed with ADHD was due to a significantly smaller left caudate nucleus and was most notable with males diagnosed with ADHD.

Brain single photon emission computed tomography (SPECT) has also indicated that children diagnosed with ADHD suffer from prefrontal cortex dysfunction. For example, Amen, Paldi, and Thisted (1993) conducted a study with 54 patients diagnosed with ADHD and a group of 18 control subjects using SPECT during a rest period and an intellectual stress period. It was found that 65% of the children diagnosed with ADHD demonstrated significant prefrontal cortex deactivation during the intellectual challange compared to only 5% of the control group.

In addition to structural abnormalities, it also has been shown that patients diagnosed with ADHD have neuro-physiological abnormalities. Zametkin, Liebenauer, Fitzgerald, and King (1993) examined the brain metabolism in a group 10 of teenagers diagnosed with ADHD and in a group of 10 normal control teenagers utilizing positron emission tomography (PET). The two groups did not differ in terms of their global measures of brain metabolism. However, the ADHD group demonstrated significantly less brain metabolism to the anterior regions of the brain compared to the control group. Interestingly, the severity of symptoms significantly correlated with diminished metabolism in the left anterior frontal lobe rather than the right anterior frontal lobe.

Similarly, Lou et al. (1989) conducted a regional cerebral blood flow study with a group of 19 children who had been diagnosed with ADHD. Six of these children had a diagnosis of only ADHD and 13 of these children had ADHD in combination with at least one other neuropsychological symptom. This study also utilized nine children without a history of psychological impairments as a control group. Results indicated that the children who had a diagnosis of ADHD or ADHD in addition to other neuropsychological impairments evidenced hypoperfusion to the striatal regions of the frontal cortex. In addition, methylphenidate appeared to increase blood flow to these regions.

Finally, Crawford and Barabasz (1996) recently examined the eletroencephalograph (EEG) magnitudes from a group of seven children with ADHD and a group of seven children without ADHD using 19 different sites. During EEG monitoring children were asked to close their eyes and listen to a story, and perform mental arithmetic with their eyes open. Results indicated that children diagnosed with ADHD evidenced lower alpha wave magnitude on the right hemisphere frontal lobe compared to the left hemisphere while listening to the story. The authors state that this data suggests the right frontal regions are less active relative to the left hemisphere in children that are diagnosed with ADHD.

Therefore, there is good reason to believe that children diagnosed with ADHD suffer from underlying prefrontal cortex dysfunction. This belief stems from the fact children diagnosed with ADHD perform in a similar fashion on neuropsychological tasks relative to patients who are known to suffer from prefrontal cortex dysfunction. In addition, the studies just described have documented prefrontal cortex dysfunction in children diagnosed with ADHD using neuroimaging and EEG techniques. Consequently, it should be expected that a pediatric medical condition that has been associated with prefrontal cortex dysfunction should also be related to the behavioral disturbances identified with ADHD.

Behavioral Inhibition and ADHD

In an attempt to create a unified theory of attention deficit hyperactivity disorder, Barkley (1997) recently proposed that the topography of cognitive and behavioral deficits associated with ADHD have their roots within the prefrontal cortex. He proposed that prefrontal cortex dysfunction leads to a primary deficit with behavioral inhibition. He also proposed that behavioral disinhibition produces secondary deficits to four other executive neuropsychological functions that are partially dependent upon behavioral inhibition for their successful execution. The four other executive functions include (1)-working memory, (2) self-regulation of affect-motivation-arousal, (3) internalization of speech, and (4) reconstitution. Barkley (1997) suggests further that behavioral inhibition in addition to these four executive functions are ultimately necessary for effective motor control, which he refers to as "motor control-fluency-syntax."

Barkley (1997) developed his unified theory of ADHD for several different reasons. First, he stated that the research examining this disorder has been primarily atheorectical and almost exclusively exploratory and descriptive in nature. The notable exceptions to this trend that Barkley (1997) noted are the Quay-Gray model (Quay, 1988; Gray, 1982) and the work of Sergeant and van der Meere (Sergeant, 1995; van der Meere, van Baal & Sergeant, 1989).

The Quay-Gray model suggested that deficits in the brain's behavioral inhibition system results in impulsiveness among children diagnosed with ADHD. This impulsiveness stems from an inability of those diagnosed with ADHD to be sensitive to environmental cues that have been associated with punishment. On the other hand, Sergeant and van der Meere (Sergeant, 1995; van der Meere, van Baal & Sergeant, 1989) proposed that the attention deficits observed in children diagnosed with ADHD are associated with information-processing impairments. While both of these theories address aspects of ADHD, neither is able to account for the entire spectrum of behavioral and cognitive deficits seen in children diagnosed with ADHD.

A second reason Barkley (1997) felt the need to create a unified theory of ADHD is that inattention, hyperactivity and impulsivity have been used to describe the disorder. However, these deficits do not seem to account for many of the other behavioral and cognitive deficits associated with ADHD such as impairments with the ability to delay gratification and the ability to develop rule-governed behaviors.

Barkley (1997) also stated that a comprehensive theory of ADHD must address several weaknesses in the literature. First, the theory must address why an actual deficit in attention has not been found (Schachar et al., 1993, 1995; Sergeant, 1995) despite the fact that parent and Teacher Rating Scales identify inattention as a primary deficit. Second, it must delineate the relationship between behavioral inhibition and attention as well as the link that these two factors have with the executive functions associated with the prefrontal cortex. A third and related requirement of the theory is that it must bridge the gap between ADHD literature and the literature of developmental neuropsychology. This gap needs to be bridged so that the relationship between ADHD and disruptions with the development of higher neuropsychological functions can be more fully understood. Finally, Barkley (1997) stated that a comprehensive theory of ADHD must be able to make specific recommendations for future research and provide predictions and methods of falsification.

<u>Origins of the Model</u> - Barkley (1997) is the first to develop a comprehensive and unifying theory of ADHD. However, much of his theory stems from the work others who have noted that the uniqueness of human language (Bronowski, 1977) and the functions served by the prefrontal cortex (Fuster, 1995, 1989; Knights, Grabowecky, & Scabini, 1995) allow for effective behavioral inhibition. Bronowski, (1977) described four unique properties of human language that enable humans to reflect upon environmental contingencies and behavioral repertoires. Additionally, these unique properties enable humans to delay or inhibit responses in accordance with environmental cues and social context. The four unique properties that are included in Bronowski (1977) theory

include: (1) prolongation, (2) separation of affect, (3) internalization of speech, and (4) reconstitution.

Prolongation refers to the human ability to relate past events with future behavior. The ability to recall past events and manipulate the imagery of these events allows for the construction of hypothetical situations and behaviors as well as the ability to predict consequences associated with hypothetical behaviors. In affect, this property allows for planning.

Separation of affect refers to the separation of an emotional event from the content of the message. This property enables humans to separate affect from motor responses. As a result, humans are able to inhibit emotionally laden responses in favor of neutral responses in the presence of emotionally charged situations.

Internalization of speech refers to the ability to turn language from a method for communicating with others into a method of reflection and thought. This property of human language functions as a self-guidance system and assists in the formation of plans and the generation of alternative responses for a given situation.

The fourth property, reconstitution, is possible through the internalization of speech. Reconstitution refers to the ability to generate new possibilities using old information. This process involves two distinct steps: First, there is an analysis of stimulus-response sequences and then a synthesis of new alternatives, wherein the former stimulus-response sequences are reconstructed in order to generate alternative behaviors.

These four unique properties taken together enable humans to inhibit immediate responses in favor of generating new, more adaptive forms of responding. Bronowski (1977) also speculated that these four unique properties of human language are possible through the functions of the prefrontal cortex.

In addition to the Bronowski (1977) model, Barkley's (1997) unifying theory of ADHD also shares a great deal with Fuster's (1995, 1989) theory. Unlike Bronowski's

(1977) model, Fuster's (1995, 1989) theory addresses the functions of the prefrontal cortex in a more direct manner. He proposed that the prefrontal cortex is essential for the generation and execution of complex and novel behaviors that bridge the temporal gap that exists between past behavioral contingencies and the current situation. Consequently, the hallmark of the prefrontal cortex is the ability to create novel behavioral sequences and inserting temporal gaps between stimulus-response-consequence chains. In addition to creating novel behavioral structures, the prefrontal cortex also is capable of creating increasingly complex behaviors from smaller behavioral units.

As a result, the prefrontal cortex is able is synthesize novel behaviors with increasing complexity and long term objectives. This is very similar to Bronowski's (1977) conceptualization of reconstitution. Additionally, Fuster (1995, 1989) proposed two other constructs that are very similar to Bronowski's prolongation construct. These constructs are called retrospective functions and prospective functions. Both of these constructs enable the prefrontal cortex to link past behavioral contingencies with present circumstances in order to generate novel behavioral sequences. These two constructs are also very similar to the neuropsychological concept of working memory. It was noted in both Fuster's (1995, 1989) theory and Bronowski's (1977) model that the inhibition of off-task information from external and internal sources is critical during the temporal delay that exists between stimulus-response sequences in which novel responses are generated. Fuster (1995, 1989) also proposed that more off-task information is able to enter working memory with ineffective inhibition. This increase in off-task information then makes it more difficult to generate novel responses. It also leads to distractibility, hyperactivity and impulsivity, all of which are associated with attention deficit hyperactivity disorder.

The theories proposed by Bronowski (1977) and Fuster (1995, 1989) have much in common and complement each other very well. Both theories note that humans have the unique ability to generate novel and complex behavioral repertoires from smaller behavioral chains by linking past behavioral contingencies with current circumstances. Bronowski (1977) placed great emphasis on the internalization of speech as the unique property of human language that enables humans to postpone responding in favor of creating new, more adaptive, responses. Fuster (1995, 1989) placed comparatively little emphasis on the role of internalized speech. However, Fuster placed much more emphasis on the ability of the prefrontal cortex to generate motivational states that give an impetus to goal-directed behavior. Although these two theories complement each other very well, neither theory addresses ADHD directly. Instead, both theories address the functions of the frontal cortex more generally. Consequently, in an attempt to create a comprehensive and unifying theory of ADHD, Barkley (1997) proposed a hybrid of these two theories.

<u>Barkley's Theory</u> - Behavioral inhibition is the executive function that is at the center of Barkley's (1997) theory. He proposed that behavioral inhibition is necessary for the successful execution of four secondary executive functions that includes (1)-working memory, (2) internalization of speech, (3) self-regulation of affect-motivation-arousal, and (4) reconstitution. It is behavioral inhibition along with these four secondary executive functions that influences goal-directed behaviors referred to as the motor control-fluency-syntax. Barkley (1997) also proposed that these executive functions originate in the prefrontal cortex and exert their influence beyond the frontal lobes into the other regulatory systems of the cerebral cortex.

The behavioral inhibition component of Barkley's (1997) model includes three types of inhibition that sets the occasion for the occurrence of the four secondary executive functions. The first type of inhibition is the ability to inhibit the execution of well-learned responses in the presence of familiar stimuli. The second type of inhibition is the ability to terminate an already ongoing response and the third type of inhibition is the ability to keep off-task information from entering working memory.

Working memory is the first of the secondary executive functions to be discussed. As previously mentioned, working memory is very similar to Bronowski's (1977) prolongation construct and Fuster's (1995, 1989) retrospective and prospective functions. All of these constructs refer to the ability to hold and manipulate multiple events and memories from the past and present in mind in order to produce hypothetical alternative behaviors that are more adaptive for the current context.

The self-regulation of affect-motivation-arousal component is very similar to the separation of affect concept described by Bronowski (1977). This secondary executive function creates the ability to self-regulate emotional states. This function also induces motivational states so that goal-directed behaviors may occur. This executive function also makes it possible to separate the decision to respond from the emotional charge that was created during a given situation. On the other hand, this function also makes it possible to generate a motivational state in the absence of an environmentally induced emotional charge.

The third executive function that is partially dependent upon behavioral inhibition in Barkley's (1997) model is the internalization of speech. This function was the central component of Bronowski's (1977) model. It is the internalization of speech that enables humans to engage in internal activities and thoughts such as the description and reflection of situations and the ability to engage in rule-governed behavior and moral reasoning.

The final executive function described in Barkley's (1997) model is referred to as reconstitution. This construct is very similar to the construct described by Bronowski (1977). Reconstitution is the ability to separate behavioral sequences into their individual parts. The individual parts of the behavioral sequences can then be re-synthesized with the individual parts of other behavioral repertoires that already exist in memory in order to create entirely novel behavioral sequences.

Consequently, inhibition sets the occasion for the execution of the four secondary executive functions. Behavioral inhibition along with these four executive functions shifts the control of motor behaviors from exclusively external sources to internally represented information. Also, during the execution of a goal-directed behavioral sequence, working memory allows for the evaluation and modification of the behavioral sequence as it occurs. Furthermore, in the event that the goal-directed behavioral sequence is interrupted, working memory enables the person to disengage from the behavioral sequence and allows for a re-engagement in the goal-directed behavior once the interruption has ceased.

<u>Support for Barkley's Theory</u> - Barkley's (1997) theory is very comprehensive. Consequently, many predictions can be made about the performance of children diagnosed with ADHD on a wide variety of tasks. Many of the predictions and hypothesis stemming from Barkley's (1997) theory are supported through the existing research findings. A cursory review of this supportive literature is presented here.

Behavioral inhibition deficits are at the center of Barkley's (1997) theory. His theory predicts that a lack of behavioral inhibition will have a detrimental impact upon the successful execution of the four secondary executive functions previously described. However, the lack of behavioral inhibition itself leads directly to behavioral impairments such as poor self-control and an inability to generate and execute goal-directed behaviors as represented by the motor control-fluency-syntax. Consequently, Barkley's (1997) model predicts that the behavior of those diagnosed with ADHD is controlled more by immediate environmental contingencies rather than internally represented environmental contingencies.

In support of this hypothesis, there have been numerous studies that have shown that children diagnosed with ADHD demonstrate significant deficits in behavioral inhibition in the presence of immediately available rewards. For example, Campell, Pierce, March, Ewing, & Szumowski (1994) found that pre-school children diagnosed with ADHD have a more difficult time delaying gratification and resisting an immediately available temptation than normal controls. Further support for Barkley's hypothesis is evidenced by the fact that researchers have found that children with ADHD have a more difficult time performing delayed response tasks (Schweitzer & Sulzer-Azaroff, 1995; Songuga-Barke, Taylor, Sembi, & Smith, 1992) and have been shown to interrupt the conversation of others more frequently (Malone & Swanson, 1993).

Behavioral inhibition deficits are also demonstrated by impaired performances on passive avoidance tasks. Such tasks require the inhibition or termination of a response in order to avoid punishment. Tasks such as these are analogous to the ability to stop or refrain from talking out of turn in a classroom setting. Indeed, researchers have found that children diagnosed with ADHD are less able to inhibit responses (Milich et al., 1994) and terminate ongoing responses following cues (Oosterlaan & Sergeant, 1998; Schachar et al., 1993; Schachar & Logan, 1990) in order to passively avoid punishing consequences.

The inability to terminate an ongoing response following a cue suggests that children with ADHD are unable to modify behavioral responses following feedback. It was suggested by Fuster (1989) that prefrontal cortex dysfunction results in the inability to modify a behavioral response following a cue. The Wisconsin Card Sorting Task (WCST) requires that subjects terminate a response pattern following a cue demonstrating its ineffectiveness. This test has been shown to be a sensitive indicator of dorsolateral prefrontal cortex dysfunction with patients that have suffered frontal lobe damage (Berman, et al., 1995). Barkley (1992) reviewed 13 studies that examined the performance of patients with ADHD on this task. He found that eight of the studies demonstrated that ADHD patients performed significantly more poorly than normal controls. Such evidence, again, suggests that children with ADHD are unable to modify behavioral responses following feedback.

Barkley's (1997) behavioral inhibition construct also predicts that patients with ADHD will be less able to inhibit off-task information. Evidence supporting this conclusion comes from studies that have utilized the Stroop Color-Word Interference Task. This task requires subjects to process a single characteristic of a stimulus (i.e. the color) while simultaneously ignoring another salient characteristic of the stimulus (i.e. the word). Barkley et al. (1992) conducted a review of six studies that utilized this test and found that all six studies showed deficient performances with patients diagnosed with ADHD. Furthermore, Leung and Connolly (1996) demonstrated that children who exhibit hyperactive tendencies are more apt to be distracted by off-task stimuli embedded within the task. Interestingly, however, these same researchers (Leung & Connolly, 1996) also found that hyperactive children are able to ignore off-task information that is not embedded within the task as well as normal controls. In sum, these reviews support the contention that children diagnosed with ADHD are less able to inhibit off-task information from entering working memory relative to normal controls.

In addition to primary behavioral inhibition deficits, Barkley's (1997) theory includes deficits associated with the four executive functions that are partially dependent upon behavioral inhibition for their successful execution. Working memory is one of the secondary executive functions partially dependent upon behavioral inhibition. As previously described, working memory is a neuropsychological construct that is very similar to Bronowski's (1977) prolongation construct and Fuster's (1995, 1989) retrospective and prospective constructs. All of these constructs involve the ability to recall and hold information in mind while simultaneously manipulating other pieces of information.

Working memory is often assessed using mental arithmetic tasks and complex memory tasks such as serial digit spans. In support of Barkley's (1997) theory, researchers (Barkley, DuPaul & McMurray, 1990; Zentall & Smith, 1993) have found that children diagnosed with ADHD are less proficient at mental arithmetic. Barkley, Murphy, and Kwasnik (1996) also found that children diagnosed with ADHD perform significantly more poorly on a backwards digit span task relative to a forward digit span task. Such a discrepancy between forward digit span and backward digit span is indicative of working memory impairments. This is due to the fact that backward serial digit spans require subjects to hold information in memory while simultaneously manipulating it. More generally, researchers (Anastopoulos, Spisto, & Maher, 1994; Lufi, Cohen, & Parish-Plass, 1990) have found that children diagnosed with ADHD score more poorly on the Freedom From Distractibility factor of the Weschler Intelligence Scale for Children - Revised.

Another one of the secondary executive functions that is partially dependent upon behavioral inhibition for its successful execution is the self-regulation of affect-motivation-arousal. As previously mentioned, this secondary executive function was initially described by Bronowski (1977) and is described as the ability to self-regulate and even induce motivational states in order to create an emotional climate for goal-directed behaviors to occur. In support of Barkley's (1997) contention that children with ADHD are unable to successfully regulate their emotional states, Pelham and Bender (1982) found that children with ADHD are more negative and emotional in their interactions with others compared to normal controls. In addition, several researchers have found a relationship between emotional control and other symptoms associated with ADHD. For example, Shoda et al. (1990) found a significant relationship between poor inhibition on a resistance to temptation task and parents ratings of emotional control in children and frustration tolerance in adolescents. Also, Eisenberg et al. (1993) found a significant relationship between negative emotional intensity and teachers' ratings of interference control.

In addition to the ability to control negative emotional intensity, the self-regulation of affect-motivation-arousal construct in Barkley's (1997) model is also responsible for creating motivational energy necessary for the completion of tasks. Support for this type of deficit in this area comes from studies in which children with ADHD have been found to be less productive with written arithmetic than normal controls (Barkley, DuPaul, et al., 1990). Studies have also shown that children with ADHD are less persistent on laboratory tasks (Barber et al., 1996; Douglas & Benezra, 1990; Wilkison, Kircher, Mcmahon, & Sloane, 1995). These studies taken together give strong support for the self-regulation of affect-motivation-arousal construct in Barkley's (1997) model.

The third executive function in Barkley's (1997) theory that is partially dependent upon behavioral inhibition for its successful execution is the internalization of speech. This construct was at the center of Bronowski's (1977) theory. It has been hypothesized (Barkley, 1997; Bronowski, 1977) that the internalization of speech enables humans to engage in internal activities such as the description and reflection of situations and the ability to engage in rule-governed behavior and moral reasoning. Only a few studies have been conducted that have directly examined this ability in children diagnosed with ADHD.

However, August (1987) conducted a study that indirectly lends support to the hypothesis that children with ADHD exhibit less internalization of speech than normal controls. This study found that children with ADHD are less likely to organize material or generate a memorization strategy individually during a memorization task.

Furthermore, August (1987) found that when these same children were given a strategy to use they typically discontinued using the rule after only a few trials. Douglas and Benezra (1990) obtained similar results. In addition, Conte and Regehr (1991) found that children diagnosed with ADHD were less able to transfer rules initially learned in one situation to a novel situation with similar environmental contingencies. Consequently, although only a few studies have indirectly examined this hypothesis, it does appear that children diagnosed with ADHD are less able to generate rules and strategies that can be used to govern their behavior.

The final secondary executive function that is partially dependent upon behavioral inhibition for its successful execution is reconstitution. As previously mentioned, reconstitution is a construct that was used by Bronowski (1977) and is described as the ability separate behavioral sequences into their individual parts. The individual parts can then be re-synthesized with the individual parts of other behavioral sequences that already exist in memory in order to create entirely novel behavioral sequences. In short, reconstitution could also be described as creativity.

Again, there are very few studies that have directly examined this ability in children diagnosed with ADHD. However, Funk, Chessare, Weaver, and Exley (1993) found that children with ADHD are less creative on a figural drawing task than normal controls. Also, Alessandri (1992) found that children with ADHD are less creative during free play than normal controls. Others studies that support this hypothesis have shown that children diagnosed with ADHD are less verbally fluent than normal controls. For example, Grodzinsky and Diamond (1992) found that children with ADHD are less proficient at a simple verbal fluency task. Also, Tannock, Purvis and Schachar (1996) found that children with ADHD are less verbally productive than normal controls during narrative stories. The poor verbal fluency observed in children diagnosed with ADHD may be the result of causes other than deficient reconstitution or creativity and more research is certainly needed in this area. However, these studies do suggest that children with ADHD may have trouble creating novel behavioral sequences.

Behavioral inhibition and the secondary executive functions that are partially dependent upon behavioral inhibition for their successful execution all enable greater control, persistence and novelty to behaviors that are goal-directed (Barkley, 1997; Fuster, 1995, 1989). The motor component in Barkley's (1997) theory is referred to as the motor control-fluency-syntax. Stemming from Barkley's (1997) theory, it should be predicted that children diagnosed with ADHD should also exhibit less motor control than normal subjects. In support of this contention, Denkla, Rudel, Chapman, and Krieger (1985) found children with attention difficulties demonstrate more neurological "soft" signs than either normal children or children with learning disabilities. Denkla et al. (1985) attributed these "soft" signs as indicators of delayed motor inhibition development. In addition, Oosterlaan & Sergeant (1998) have conducted research that suggests the motor control deficit seen in children diagnosed with ADHD comes from an inability to prepare for motor action rather than from an inability to maintain attention or choose appropriate responses.

Behavior Rating Scales

As described previously, researchers frequently have used neuropsychological tests to identify many of the behavioral and cognitive impairments associated with ADHD. In addition to these tests, however, standardized behavior rating scales have gained wide respect for the assessment of childhood behavior disorders (Gross and Wixted, 1988). In fact, Barkley et al. (1990) stated that standardized behavior rating scales are the second most important tools used for assessing and diagnosing ADHD. Standardized behavior rating scales make it possible for adults to objectively quantify their opinions concerning the behavior of individuals in their care. It is also possible to generate standard scores from these quantified opinions and compare these scores with age and gender matched normative samples.

Several problems are inherent when using and interpreting standardized behavior rating scales. Rating scales are structured, quantified opinions and are subject to response bias. For instance, Abikoff, Courtney, Pelham, and Koplewicz (1993) found that rating scales are vulnerable to “halo effects,” which is the rater’s tendency to overreport ADHD symptoms in children who are difficult and oppositional. In addition, it is assumed that the rater is able to extract relevant behaviors across a wide variety of settings and situations and accurately reflect these behaviors on the rating scale (Reid and Maag, 1994).

Despite these limitations, however, standardized behavior rating scales offer many advantages for both research and clinical endeavors. Standardized behavior rating scales frequently have normative data. Such data makes it possible for the clinician or researcher to determine the statistical deviance of a behavior pattern compared to a normal population of age and gender matched children. This characteristic is especially beneficial when assessing a child for ADHD. Many of the behaviors associated with ADHD occur in children as a part of normal childhood development. Consequently, it is necessary to compare the behavior pattern of the child being assessed with the behavior patterns of a normative sample that is matched in terms of age and gender in order to establish determine if the behavior pattern is truly deviant (Barkley et al., 1990).

Second, behavior rating scales make it is possible to gather information concerning the child’s behavior from an informer that has had many interactions with the child in a wide variety of settings. This makes it is possible to quickly assess behaviors that occur infrequently and are unlikely to be observed during direct behavioral observation. This attribute also makes it possible to filter out situational variability in favor of focusing upon the most stable behavioral characteristics of the child (Barkley et al., 1990).

In fact, Barkley (1994) has stated that the assessment of these stable characteristics is the most reliable and valid indicator of behavioral disturbances associated with ADHD. In addition, Barkley (1994) stated that it is unlikely that the behavioral disturbances associated with ADHD will be reliably detected by neuropsychological assessments that occur during brief periods of time. Instead, the disturbances associated with ADHD will most likely emerge from an analysis of how the person functions in response to everyday demands. These views stem from the fact that ADHD is a dysregulatory phenomenon rather than a specific skill deficit. As a result, the observations made by others collapsed across time may be much more valid indicators of the presence of ADHD than traditional neuropsychological assessments can provide (Barkley, 1994).

Standardized behavior ratings also make it possible to quantify and assess the qualitative aspects of behavior. Many of the disruptive behaviors exhibited by children are highly subjective and difficult to quantify through direct behavioral observation or neuropsychological testing. In addition, it is often these qualitative aspects of behavior that are most salient to the child's caregivers. Furthermore, caregivers frequently base their responses toward their child on these qualitative aspects. Consequently, it is extremely important to understand and quantify the subjective perceptions that caregivers have concerning their child's behavior patterns so that these perceptions can be compared to the views of caregivers who interact with normal children (Barkley, 1990).

Fourth, standardized behavior rating scales provide a means for assessing the clinical efficacy of various treatment plans (Barkley et al., 1990). This may not be the case for most of the neuropsychological tasks used to assess ADHD. While neuropsychological tasks may assess more enduring deficits associated with ADHD, parent and Teacher Rating Scales afford the researcher and clinician the opportunity to assess changes in the child's ability to adapt and function in the everyday environment.

Finally, Power and Ikeda (1996) recently conceded the fact that rating scales used in isolation may lead to incorrect conclusions regarding the presence of ADHD. However, these authors along with others (Gross and Wixted, 1988) state that behavior-rating scales serve an invaluable function during the initial screening of problem behaviors. Such a screening allows the clinician or researcher to identify children who exhibit many of the behaviors associated with ADHD so that they may be evaluated in a more comprehensive manner in the future.

<u>Behavior Assessment System for Children</u> - The Behavior Assessment System for Children (BASC) (Reynolds and Kamphaus, 1992) is one standardized behavior rating scale that has gained popularity and respect in recent years. This is a system of five parts that consists of both Parent and Teacher Rating Scales, a student observation system, a developmental history, as well as a self-report measure. Both the style and structure of the parent and teacher rating forms are very similar.

The Parent Rating and Teacher Rating Scales are available in three different forms: pre-school (ages 4-5), child (ages 6-11), and adolescent (ages 12-18). The different forms are designed to reflect different developmental stages of behavior. All of the forms measure behavior on a 4-point scale of frequency from (1) never to (4) almost always and assess the child's adaptive and problem behaviors at home and at school as well as in social situations. Each form takes approximately 20 minutes to complete.

The clinical scales of the Parent Rating Scale consist of Aggression, Anxiety, Attention Problems, Atypicality, Conduct Problems, Depression, Hyperactivity, Learning Problems, Somatization, and Withdrawal. The rating form also consists of several composite scales. The Externalizing Problems composite consists of the Hyperactivity, Aggression, and Conduct Problems clinical scales. The Internalizing Problems composite consists of the Anxiety, Depression, and Somatization clinical scales. A Behavior Symptoms Index consists of the scores from the Hyperactivity, Aggression, Anxiety, Depression, Attention Problems, and Atypicality clinical scales.

The Teacher Rating Scale contains all of the clinical scales and composite scales included as part of the Parent Rating Scale. It also contains two additional clinical scales including the Learning Problems and the Study Skills clinical scales. The Teacher Rating Scale also includes a School Problems composite that consists of the Attention Problems and Learning Problems clinical scales.

The Parent rating and teacher rating forms of the BASC also contain scales that are designed to measure positive behaviors. The Parent rating form includes Adaptability, Leadership, and Social Skills scales. All three of these scales are combined to arrive at the Adaptive Skills composite. The Teacher Rating Scale uses these three scales in addition to the Study Skills scale to arrive at the Adaptive Skills composite. The Parent rating and teacher rating forms also contain an F index designed to indicate the tendency of the rater to judge the behavior of the child in an unusually negative manner.

Both rating forms yield T-scores and percentiles for each clinical and composite scale based upon a national normative sample. Reynolds and Kamphaus (1992) suggest the following T- score cutoffs to aid with interpretation: 70+ Clinically Significant; 60-69 At-Risk; 41-59 Average; 31-40 Low; and -30 Very Low. The normative sample for the Parent rating form consists of 3,483 children from across the United States at 116 testing sites. The normative sample for the teacher rating form consists of 2,401 children tested at the same sites. At each site, two classrooms per grade and four randomly selected children per classroom were selected for both the parent and teacher ratings. This normative data is grouped by gender and age in order to allow comparisons of childhood behaviors in relation to a normal sample that has been age and gender matched.

Weighting also was done in order to match the normative sample with the general population in terms of maternal education, geographic region, and special education placement. It is also noted that the normative sample closely matched the general population so the weighting process probably did not significantly distort the final normative data (Reynolds and Kamphaus, 1992). The authors also included normative data on several groups of emotionally and behaviorally disturbed children including those diagnosed with conduct disorder (n=40), depression (n=29), autism (16), and attention deficit hyperactivity disorder (n=52).

The authors (Reynolds and Kamphaus, 1992) report average internal consistency reliabilities of .80. It is also noted that these reliabilities generally increase with age until consistencies of nearly .90 are achieved for adolescents. These reliabilities are also similar across gender and are highest for the externalizing and adaptive rating scales. The internal consistency reliabilities are also very similar between the normal sample and the clinical samples.

Both the Parent rating and the teacher rating forms also exhibit satisfactory test-retest reliabilities. This type of reliability was also typically in the middle .80s to the middle .90s over a one-month period for both forms. Again, reliabilities tend to increase with age. In addition, the authors (Reynolds and Kamphaus, 1992) conducted a seven month test-retest reliability study with a clinical sample of young children and found a median correlation of .69 suggesting stability over a longer period of time.

Reynolds and Kamphaus (1992) also report concurrent validity with three of the most commonly used Parent Rating Scales. The Parent Rating Scale correlated between .70 and .80 with the Child Behavior Checklist and the Personality Inventory for Children - Revised on scales designed to measure similar constructs. In addition, both the

Hyperactivity and Externalizing scales of the Parent Rating Scale correlated .56 with the Conners Hyperactivity Index. The correlations between the teacher rating form and the Achenbach scales are also quite high, ranging in the .80s and .90s. The validity of the BASC is also supported in terms of the performance of several groups of children diagnosed with psychiatric disorders including those diagnosed with attention deficit hyperactivity disorder.

The inter-rater reliabilities for the teacher rating form demonstrate reasonably high correlations ranging from .69 on the Internalizing Problems composite to .89 on the School Problems composite. However, the inter-rater reliabilities between the ratings of parents is considerably lower. The median correlations for the preschool, child, and adolescent forms are respectively .46, .57, and .67. The lowest values were obtained for the Internalizing Problems composite for all three age groups. This discrepancy between parent ratings has been found frequently in other studies examining correlations between parents' ratings of childhood behaviors.

Reynolds and Kamphaus (1992) collected normative data on 52 children that had been diagnosed with ADHD. This sample of children demonstrated the highest elevations on the Hyperactivity (T = 68.0, SD = 14.5) and Attention Problems (T = 65.7, SD = 7.4) clinical scales when using the parent rating form. This group also demonstrated significantly lower scores on the Adaptability (T = 39.0, SD = 10.1) composite scale when using the parent rating form. When using the teacher rating form, the children diagnosed with ADHD evidenced very similar profiles when compared to the profiles obtained when using the parent rating form. Although, the high points on the parent rating form are more severe and the Adaptability composite scores are lower. Consequently, the profiles obtained using both the parent and teacher rating forms for this clinical sample are consistent with the disturbances associated with ADHD. It also appears that the Parent Rating Scale differentiate children with ADHD better than the

Teacher Rating Scale. Manning-McGallian and Miller (1997) compared the PRS profiles of the BASC (Reynolds and Kamphaus, 1992) between 40 children diagnosed with ADHD and 31 normal controls. Similar to the study conducted by Reynolds and Kamphaus (1992), these researchers found extremely significant ($p < .001$) differences between the children diagnosed with ADHD and the normal controls on the Attention and Hyperactive clinical scales as well as on the Adaptive Skills composite scale. More recently, Ostrander et al. (1998) utilized the BASC and the Child Behavior Checklist (CBCL) in order to differentiate ADHD and non-ADHD among school children. This study found that the BASC was more parsimonious and accurate than CBCL at differentiating ADHD from non-ADHD students.

Attention Deficit Disorders Evaluation Scale - The Attention Deficit Disorders Evaluation Scale (ADDES) was developed and designed by McCarney (1995) to provide a direct measure of the characteristics that have been used define ADHD by The American Psychiatric Association: Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV, 1994). This measure contains two versions including the home version and the school version in order to provide examiners the ability to assess the presence of ADHD in more than one setting as recommended by the DSM-IV (1994). The primary care giver or legal gaurdian serves as the rater when using the home version and the child's primary teacher serves as the rater when using the school version. The home version contains 46 items and the school version contains 60 items. Both forms measure behavior on a 5-point scale of frequency from (0) Does Not Engage In The Behavior; to (4) Engages In The Behavior One To Several Times Per Hour. Each form takes approximately 10 minutes to complete.

Both versions contain two subcales including the (1) inattention and, (2) hyperactivity-impulsivity subscales. These two subscales provide scaled scores and were developed from a factor analysis of all the items comprising the scale as well as face validity based upon the DSM-IV (1994) groupings. Both versions also include a percentile score that is intended to provide a global measure of behaviors that constitute ADHD in reference to a standardization sample.

The home version utilizes a standardization sample of 2,415 children and youth between the ages of three and twenty from 23 states. Ten standardization groups were developed from this normative sample in order to control for age and gender differences. The school version utilizes a standardization sample of 5,975 students between the ages of four and nineteen years from 30 states. Again, ten standardization groups were developed from this normative sample in order to control for age and gender differences.

The ADDES-Home Version and the ADDES-School Version also demonstrate adequate psychometric properties. The test-retest coefficients for the home version range from .88 to .93 when parents were asked to complete the ADDES 30 days following their first rating. Similarly, the test-retest coefficients for the school version range from .88 to .97 when teachers were asked to complete the ADDES 30 days from their first rating. McCarney (1995) also reports the inter-rater reliabilities of both the home version and the school version. The inter-rater reliabilities of the home version range from .80 to .84 and the inter-rater reliabilities of the school version range from .81 to .90.

In order to establish diagnostic validity for the ADDES - Home Version, McCarney (1995) compared subscale scores and percentile scores between 102 children that were randomly selected from the normative sample and 102 children diagnosed with ADHD. Each group included 72 males and 30 females and represented children between the ages of 3.0 and 19.0 years. Psychologists or pediatricians outside of the school system diagnosed all of the children. Results demonstrated that both males and females diagnosed with ADHD obtained significantly lower subscale and percentile scores compared to the subscale and percentile scores obtained by their same sex counterparts in the normative sample ($p<.001$). These low subscale and percentile scores indicate that the children diagnosed with ADHD demonstrate significantly more behaviors that are characteristic of ADHD compared to a group of normal controls.

McCarney (1995) also compared subscale scores and percentile scores between 102 children that were randomly selected from the normative sample and 102 children diagnosed with ADHD using the ADDES - School Version. Each group included 72 males and 30 females and represented children between the ages of 4.5 and 19.0 years. Again, results demonstrated that both males and females diagnosed with ADHD obtained significantly lower subscale and percentile scores compared to the subscale and percentile scores obtained by their same sex counterparts in the normative sample ($p<.001$).

CHAPTER 2

PURPOSE

The metopic suture of the human cranium lies immediately superior to the prefrontal cortex. Metopic synostosis is a condition in which the metopic suture fuses prematurely and may be associated with underlying dysfunction of the prefrontal cortex. This dysfunction may stem from increased cranial pressure due to anterior fossa constraint or primary hypoplasia of the prefrontal cortex.

It also has been suggested that the prefrontal cortex is the cerebral region responsible for behavioral inhibition and executive function and is often compromised in children diagnosed with ADHD (Barkely, 1997). Therefore, it is reasonable to conclude that children with a history of metopic synostosis will evidence many of the same behavioral disturbances associated with ADHD. However, no study has examined the presence of behavioral disturbances in children with a history of metopic synostosis.

The purpose of the current project is to examine the presence of behavioral disturbances in children with a history of metopic synostosis. It is postulated that children with a history of metopic synostosis will evidence many of the same behavioral disturbances associated with ADHD. It also is postulated that metopic synostosis will have a differential impact upon behavior compared to other forms of craniosynostosis.

In order to examine this phenomenon the behavior of three groups of children will be compared. The three groups of children will include a group of children with a history of metopic synostosis, a group of children that has been diagnosed with ADHD, and a group of children with a history of isolated craniosynostosis not involving the metopic suture. Behavioral comparisons will be made using the Parent rating and Teacher Rating Scales from the Behavioral Assessment System for Children (Reynolds and Kamphaus,

1992) as well as the ADDES-home version and the ADDES-school version (McCarney, 1995).

Hypotheses

Five hypotheses will be made following the purpose and intent of the current project. The first four hypotheses concern the comparisons that will be made using the BASC Parent rating and Teacher Rating Scales (Reynolds and Kamphaus, 1992). Hypothesis 1 states that a group of children with a history of metopic synostosis will evidence a behavior profile that is similar compared to a group of children diagnosed with ADHD as measured by the BASC Parent Rating Scale (Reynolds and Kamphaus, 1992). Hyothesis 2 states that a group of children with a history of metopic synostosis will evidence a behavior profile that is similar compared a group of children diagnosed with ADHD as measured by the BASC Teacher Rating Scale (Reynolds and Kamphaus, 1992). Hypothesis 3 states that the group of children with a history metopic synostosis will evidence a behavior profile that is dissimilar compared to a group of children with a history of isolated craniosynostosis not involving the metopic suture as measured by the BASC Parent Rating Scale (Reynolds and Kamphaus, 1992). Hypothesis 4 states that a group of children with a history metopic synostosis will evidence a behavior profile that is dissimilar compared to a group of children with a history of isolated craniosynostosis not involving the metopic suture as measured by the BASC Teacher Rating Scale (Reynolds and Kamphaus, 1992).

The final hypothesis concerns the comparison that will be made between the group of children with a history of metopic synostosis and the group of children with a history of saggital synostosis using the ADDES-home version and the ADDES-school version (McCarney, 1995). Hypothesis 5 states that the group of children with a history of metopic synostosis will evidence more behaviors associated with ADHD than the group of children with a history of isolated craniosynostosis not involving the metopic suture as measured by the ADDES-home version and the ADDES-school version (McCarney, 1995).

CHAPTER 3

METHOD

Participants

Three groups of children were utilized as participants for the present study. Group 1 consisted of 30 children (15 males, 15 females) who had a history of isolated metopic craniosynostosis. The ages of these children ranged from 4 years, 7 months -to 11 years, 5 months (M= 7.63 years, SD = 1.92 years). Twenty-four of these subjects had at least one craniofacial surgery while six never had craniofacial surgery. The average age of the first craniofacial surgery was 9.04 months (SD = 7.78 months) with a range from 2 months, –to 40 months. The mothers of these children reported disabilities and impairments utilizing a brief developmental questionnaire. Eleven of these children were reported to be in ‘special education,’ 12 were reported as having a ‘learning disability,’ 2 were reported to have a ‘visual impairment,’ 3 were reported having a ‘hearing impairment,’ 9 were reported having a ‘language impairment,’ and 10 were reported as having a ‘behavioral disturbance.’

Group 2 consisted of children who had previously been diagnosed with ADHD. The data for these two groups of children was collected during the initial standardization of the BASC. The data from these two groups of children was not used to establish the normative data of the BASC, but rather to demonstrate how a clinical sample of children diagnosed with ADHD compare to the normative sample of the BASC. The data for this ADHD clinical sample was obtained for the purposes of this study by contacting American Guidance Service, Inc. (AGS). The ADHD clinical sample for the BASC Parent Rating Scale consisted of 45 children and the ADHD clinical sample for the BASC Teacher Rating Scale consisted of 68 children. The ages of the BASC PRS subjects ranged from 4 years, 3 months –to 11 years, 6 months (11 males, 34 females). The average age of these subjects was 7.82 years (SD = 1.93 years). The ages of the BASC TRS subjects ranged from 5 years, 4 months –to 11 years, 10 months (22 males, 46 females). The average age of these subjects was 7.77 years (SD = 1.85 years).

Group 3 consisted of 30 children (15 males, 15 females) who had a history of isolated craniosynostosis not involving the metopic suture. Subjects within this group were age and sex matched to subjects within Group 1. The ages of these children ranged from 4 years, 0 months -to 11 years, 3 months (M= 7.54 years, SD = 1.88 years). Twenty-nine of these subjects had at least one craniofacial surgery while only one never had craniofacial surgery. The average age of the first craniofacial surgery was 9.62 months (SD = 8.53 months) with a range from 2 months –to 36 months. The mothers of these children also reported disabilities and impairments utilizing a brief developmental questionnaire. Four of these children were reported to be in 'special education,' 4 were reported as having a 'learning disability,' 4 were reported to have a 'visual impairment,' 2 were reported having a 'hearing impairment,' 5 were reported having a 'language impairment,' and 5 were reported as having a 'behavioral disturbance.'

Subjects for Group 1 and Group 3 consisted of patients that had traveled to one of two major craniofacial centers for treatment during the past twelve years. Requirements for participation included subjects that were currently between the ages of 4 years, 0 months and 11 years, 11 months and had a history simple craniosynostosis. No individual cases were used in either of these two groups in which craniosynostosis was determined to be a part of a larger syndrome or the result of another disease process.

Procedure

Subjects for Group 1 and Group 3 were recruited from the Dallas Craniofacial Center in Dallas, Texas and the Southwest Craniofacial Center in Phoenix, Arizona. An archival medical records search was conducted at these craniofacial centers in order to identify patients that met the requirements of the examination. The names, addresses and phone numbers of the patients meeting the requirements were given to the primary investigator of the study. In cases where the addresses and phone numbers were out of date an Internet based locating service was utilized in order to obtain current addresses and phone numbers.

Once this information was collected, the parents of children with a history of metopic synostosis (Group 1) were contacted by telephone. The purpose and intent of the research project was described and their participation was solicited. In the event that the parents agreed to participate, a letter, a Parent Informed Consent form, a BASC Parent Rating Scale (Reynolds and Kamphaus, 1992), the ADDES – Home Version (McCarney, 1995) and a brief developmental questionnaire were sent directly to them. The mothers were asked to complete both rating forms and the developmental questionnaire and return it along with the signed informed consent to the primary investigator within two weeks in a self-addressed envelope that was provided. The parents of these children were also provided a medium-sized enveloped that contained a letter for the child's schoolteacher, a Teacher Informed Consent Form, a BASC Teacher Rating Scale and the ADDES – School Version. They were asked to take this envelope to the child's schoolteacher and ask him or her to complete both rating forms and return them along with the signed informed consent form to the primary investigator within two weeks in a self-addressed envelope that was provided.

A total of 47 parents of children with a history of metopic synostosis were contacted by telephone. Six of these parents declined to participate upon learning the description and intent of the investigation. All of the parents that decided to participate agreed to only allow the mothers of the children to complete the questionnaires in order to control any response pattern differences that may exist between mothers and fathers.

Forty-one packets were mailed to the parents who agreed to participate and thirty packets that were at least partially complete were returned to the primary investigator. Twenty of the respondents completed all questionnaires, while 10 of the respondents agreed to complete only the parent rating portion of the study and declined to complete Teacher Rating Scales. The most frequently cited reason for not wanting to participate in the teacher rating portion of the examination was fear of stigmatizing their child (8 parents) followed by refusal on the part of the child's schoolteacher (2 teachers).

The parents of children with a history of craniosynostosis not involving the metopic suture (Group 3) were contacted by telephone after the parents of children with a history of metopic synostosis agreed to participate. This sequence of contacting participants was used so that children within Group 3 could be age and sex matched to children within Group 1. A total of 49 parents of children with a history of craniosynostosis not involving the metopic suture were contacted by telephone. Eight of these parents declined to participate upon learning the description and intent of the investigation. All of the parents that decided to participate agreed to only allow the mothers of the children to complete the questionnaires in order to control any response pattern differences that may exist between mothers and fathers.

Forty-one packets were mailed to the parents who agreed to participate and thirty packets that were at least partially complete were returned to the primary investigator. This group contained 19 cases of isolated saggital synostosis, 10 cases of isolated unilateral coronal synostosis and one case of isolated unilateral lamdoid synostosis. Twenty-five of the respondents completed all questionnaires, while 5 of the respondents agreed to complete only the parent rating portion of the study and declined to complete Teacher Rating Scales. Fear of stigmatizing their child was the reason that all five of these parents stated that they declined to complete the teacher-rating portion of the examination.

Ethical Concerns

Participants were informed about the purpose and the nature of the study upon making telephone contact. Participants were given informed consent forms, which they were asked to read and sign before initiating their participation. Participants were free to ask questions regarding the study at any time utilizing the phone numbers of the primary investigators listed at the top of the informed consent form. In addition, they were informed that they were free to withdraw from the study at anytime without consequence.

Participants were also informed that all of the collected data was coded and their names were not associated with the data. The data will be kept in a locked file and will be destroyed five years after its collection. The only people with access to the data are the primary investigators of the study.

No deception was used in this study. The only adverse effect that was foreseeable is that the parents of the children may have become sensitized to any behavioral disturbances that they might have. This was dealt with by giving the subjects the contact numbers of the researchers in case that they may have any concern about their performance in this study.

CHAPTER 4

RESULTS

The analysis of data was performed using the Statistical Package for the Social Sciences, Version 9.0 (SPSS 9.0). Results for the BASC Parent Rating Scale and the BASC Teacher Rating Scale are reported in terms of standardized T-scores (M=50, SD=10) and are based upon the sex-referenced normative data of these two scales. The sex-referenced normative data was utilized in order to control for behavioral differences between the sexes. Results for subscales of the ADDES Home and School Versions are reported in terms of sex-referenced scaled scores (M=10, SD=3.0). Results for the Total scores of the ADDES Home and School Versions are reported in terms of percentile ranks. The dependent variables are expressed in terms of standard scores, scaled scores and percentiles as indicated by the tests that were utilized in this study.

Tests of Hypothesis

Profile analyses were conducted in order to test Hypothesis 1 through Hypothesis 4. More specifically, tests of parallelism were conducted for each of the Hypothesis 1 through Hypothesis 4. Each test of parallelism consisted of two parts. First, an examination was conducted in order to determine if group profile plots are parallel to one another. That is, the similarity of shape between group profiles was examined by comparing group subscale or composite scale means. In order to satisfy parallelism of group profiles, the two profile plots needed to be parallel to each other for each line segment between adjacent subscales or composite scales. In order to complete this test, the group mean and group variance of each slope between adjacent scales is calculated. The variate of group mean and group variance is established and then compared to the group mean and variance of the comparison group thereby establishing an F ratio. The null hypothesis stated that the two profiles were similar to one another and no actual differences existed between the two profiles. The null hypothesis was rejected if $p \leq .05$ (the two profiles were not parallel to one another – they were dissimilar).

Next, if the null hypothesis was not rejected (group profiles were deemed to possess a similar shape) a test of equality was subsequently performed in order to examine the equality of the group subscales or composite scales. The variate of group scale means and variance is established and then compared to the group scale means and variance of the comparison group. The null hypothesis stated that group subscale or composite scale means were equal to one another. The null hypothesis was rejected if $p \leq .05$ (the group subscale or composite scale means are not equal to one another).

Group profiles for the BASC Parent Rating Scale are visually illustrated in Figure 1 (subscales) and in Figure 2 (composite scales). Group subscale means and standard deviations for the BASC Parent Rating Scale are listed in Table 1 (subscales) and in Table 2 (composite scales). Group profiles for the BASC Teacher Rating Scale are visually illustrated in Figure 3 (subscales) and Figure 4 (composite scales). Group subscale means and standard deviations for the BASC Parent Rating Scale are listed in Table 1 (subscales) and in Table 2 (composite scales).

Hypothesis 5 was examined using a MANOVA in order to test group mean differences of the subscales of the ADDES Home and School Versions as well as group mean differences of the Total scores of the ADDES Home and School Versions. The null hypothesis stated that no actual differences existed between the group mean differences on the subscales and Total scores of the ADDES Home and School Versions. The null hypothesis was rejected if $p \leq .05$ (group mean differences on the subscales and Total scores of the ADDES were not equal to each other). An ADHD control group was not available for this comparison. Consequently, the only comparisons that were made for Hypothesis 5 were between children with a history of metopic synostosis and children with a history of craniosynostosis not involving the metopic suture. Group profiles for the ADDES Home and School versions are visually illustrated in Figure 5 (subscales) and in Figure 6 (Total Scores). Group means and standard deviations for the ADDES Home and School versions are listed in Table 5 (subscales) and in Table 6 (Total scores).

<u>Hypothesis 1</u> - Hypothesis 1 stated that children with a history of isolated metopic synostosis would evidence a similar behavior pattern compared to a group of children diagnosed with ADHD as measured by the BASC Parent Rating Scale. Utilizing a test of parallelism, it was found that that group subscales of the BASC Parent Rating Scale differed significantly between these two groups of children, $F(11,56) = 2.285$, $p \leq .05$. The profiles were not similar between these groups. It also was found that the profile of composite scales of the BASC Parent Rating Scale differed significantly between these two groups of children, $F(3,64) = 6.11$, $p \leq .01$. Again, the profiles were not similar between these two groups.

Consequently, the results of this study do not support Hypothesis 1. Instead, the BASC Parent Rating Scale behavioral profiles of children with a history of isolated metopic synostosis are not similar (not parallel) to the behavior profiles of the children who had been diagnosed with ADHD. This lack of similarity is true for both the subscales as well as the composite scales of the BASC Parent Rating Scale. These results indicate that the mothers of children with a history of metopic synostosis perceive the behavioral patterns of their children in a dissimilar fashion relative to the manner in which mothers of children diagnosed with ADHD perceive their children's behavior.

Hypothesis 2 - Hypothesis 2 stated that children with a history of metopic synostosis would evidence a similar behavior pattern compared to a group of children diagnosed with ADHD as measured by the BASC Teacher Rating Scale. A test of parallelism was again conducted and it was found that group subscales of the BASC Teacher Rating Scale did not significantly differ between these two groups of children $F(13,70) = 1.745$, $p \geq .05$. In order to further assess the similarity of these subscale profiles a test of equality was performed next. Utilizing this test, it again was found that there was no significant difference between group subscales between these two groups of children in terms of profile equality, $F(13,70) = 2.067$, $p \geq .05$. The profiles were both parallel and equal between these two groups of children.

A test of parallelism also was performed in order to assess the similarity of the profiles of the composite scales of the BASC Teacher Rating Scale. It was found that the composite scale profiles did not differ significantly between these two groups of children, $F(4,79) = 1.132$, $p \geq .05$. Again, in order to further assess the similarity of these composite scale profiles a test of equality was performed next. Utilizing this test, it was found that there was no significant difference between the profiles of the composite scales between these two groups of children in terms of profile equality, $F(4,79) = 2.757$, $p \geq .05$. Again, the profiles were both parallel and equal between these two groups of children.

The results of this study do support Hypothesis 2. The children with a history of metopic synostosis evidence a parallel and equal behavior pattern relative to a group of children diagnosed with ADHD as measured by the BASC Teacher Rating Scale. The presence of similarity of behavior profiles between these two is true for both the subscales as well as the composite scales. These results indicate that teachers of children who have a history of metopic synostosis perceive the behavior patterns of these students in a very similar fashion relative to the manner in which teachers perceive the behavior patterns of students diagnosed with ADHD.

<u>Hypothesis 3</u> - Hypothesis 3 stated children with a history of isolated craniosynostosis not involving the metopic suture would evidence a behavior pattern that is dissimilar compared to children with a history of metopic synostosis as measured by the BASC Parent Rating Scale. A test of parallelism was conducted and it was found that the subscale profiles of these two groups of children were not significantly different from one another $F(11,34) = 1.57$, $p \geq .05$. In order to further assess the similarity of these subscale profiles a test of equality was performed next. Utilizing this test, it again was found that there was no significant difference between the profiles of the subscales between these two groups of children in terms of profile equality, $F(11,34) = 1.231$, $p \geq .05$. The subscale profiles were both parallel and equal between these two groups of children.

Another test of parallelism was performed in order to assess the composite scale similarity between these two groups of children. It was found that the composite scale profiles did not differ significantly between these two groups $F(3,56) = .724$, $p \geq .05$. Again, in order to further assess the similarity of these composite scale profiles a test of equality was performed next. Utilizing this test, it was found that there was no significant difference between the profiles of the composite scales between these two groups of children in terms of profile equality, $F(3,56) = 2.211$, $p \geq .05$. The composite profiles were both parallel and equal between these two groups of children.

These results do not support Hypothesis 3. The children with a history of metopic synostosis evidence a similar behavior pattern relative to children with a history of isolated craniosynostosis not involving the metopic suture as measured by the BASC Parent Rating Scale. The presence of similarity of behavior profiles between these two groups is true for both individual subscales as well as the composite subscales. These results indicate that the mothers of these two groups of children do not perceive the behavior patterns of these children in a dissimilar fashion from one another.

Hypothesis 4 - Hypothesis 4 stated that children with a history of isolated craniosynostosis not involving the metopic suture would evidence a behavior pattern that is dissimilar compared to children with a history of metopic synostosis as measured by the BASC Teacher Rating Scale. A test of parallelism was conducted and it was found that the subscale profiles of these two groups of children were not significantly different from one another $F(13,23) = 1.90$, $p \geq .05$. In order to further assess the similarity of these subscale profiles a test of equality was performed next. Utilizing this test, it was found that there was a significant subscale profile difference between these two groups of children in terms of profile equality, $F(13,23) = 7.361$, $p \leq .05$. Therefore, the group subscale profiles were parallel to each other, but were not equal. Another test of parallelism was performed in order to assess the composite scale similarity between these two groups of children. It was found that the composite scale profiles did differ significantly between these two groups $F(4,34) = .3.422$, $p \leq .05$. The group composite profiles were not parallel.

The results of this study partially support Hypothesis 4. The subscale profiles were parallel between these two groups although the subscales were not equal. Additionally, the composite scales were not parallel between these two groups. The dissimilarity of composite scales between these two groups indicates teachers perceive the behavior pattern of children with a history of metopic synostosis differently relative to the manner in which teachers perceive the behavior patterns of children with a history of isolated craniosynostosis not involving the metopic suture.

Hypothesis 5 - Hypothesis 5 stated that children with a history of metopic synostosis would demonstrate more behaviors associated with ADHD than children with a history of isolated craniosynostosis not involving the metopic suture as measured by the ADDES Home and School versions. Subjects within Group 1 and Group 2 were matched in terms of age and sex for the purposes of these analyses. A MANOVA was conducted to compare the subscale values of the ADDES Home and School Versions. It was found that mothers and teachers rated children with a history of metopic synostosis as engaging in more behaviors associated with ADHD than children with a history of isolated craniosynostosis not involving the metopic suture $F(4,13) = 6.82$, $p \leq .01$. A MANOVA was also conducted to compare the Total Score values for the ADDES Home and School Versions. Again, it was found that mothers and teachers rated children with a history of metopic synostosis as engaging in more behaviors associated with ADHD than children with a history of isolated craniosynostosis not involving the metopic suture $F(2,15) = 4.44$, $p \leq .05$.

The results of this study support Hypothesis 5. Hypothesis 5 utilized the ADDES Home and School Versions to compare the frequency at which behaviors associated with ADHD occur in children with a history of metopic synostosis relative to children with a history of isolated craniosynostosis not involving the metopic suture. Results demonstrated that children with a history of metopic synostosis were rated as engaging in more behaviors associated with ADHD than children with a history of isolated craniosynostosis not involving the metopic suture on both the Home Version and School Version. That is, both mothers and schoolteachers were found to perceive children with a history of metopic synostosis as possessing more ADHD qualities than children with a history of isolated craniosynostosis not involving the metopic suture.

This trend was found for both the Total scores of the ADDES Home and School versions, the Inattentiveness and Hyperactivity subscales of the ADDES School Version and the Inattentiveness subscale of the ADDES Home Version. The only test result that did not support Hypothesis 5 was the one that indicated no significant difference existed between the two groups of children on the Hyperactivity subscale of the ADDES Home Version.

Table 1.
Means of BASC Parent Rating Scale subscales with three groups of children

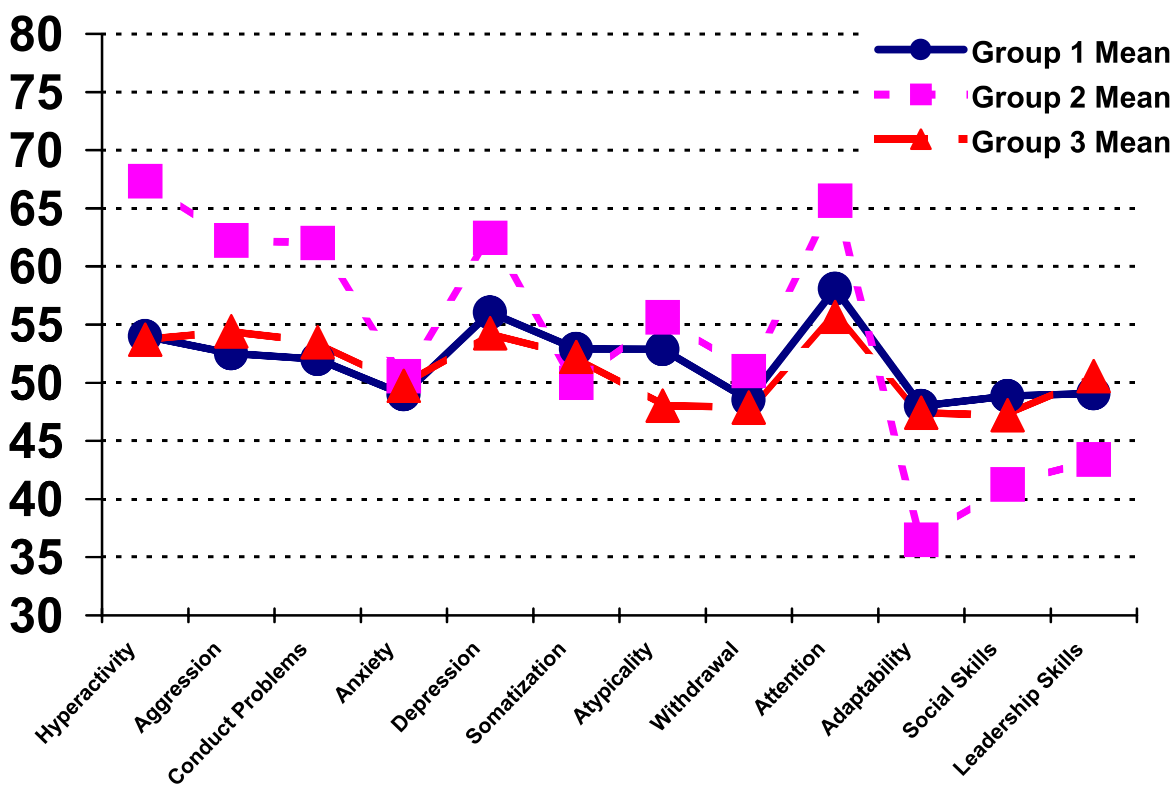
Subscale	**Group 1**		**Group 2**		**Group 3**	
	Mean	Std. Dev.	Mean	Std. Dev.	Mean	Std. Dev.
Hyperactivity	54.00	13.94	*67.33	14.98	53.70	13.41
Aggression	52.52	13.47	*62.24	12.60	54.43	12.60
Conduct Problems	52.04	11.87	*62.02	12.72	53.48	11.46
Anxiety	49.00	9.96	50.58	10.36	49.74	9.44
Depression	56.04	11.17	*62.47	12.83	54.26	13.01
Somatization	52.91	8.34	49.98	11.79	52.22	8.40
Atypicality	52.87	14.74	55.64	9.84	48.04	9.87
Withdrawal	48.52	8.90	51.00	13.22	47.87	9.31
Attention	58.08	12.25	*65.66	7.47	55.65	8.23
Adaptability	48.00	12.45	*36.49	11.93	47.43	9.05
Social Skills	48.87	9.02	*41.27	9.03	47.21	8.11
Leadership Skills	49.08	8.55	*43.40	9.60	50.57	9.20

*Significantly different from Group 1 ($p \leq .05$)

Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture



Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture

Figure 1. Means of BASC Parent Rating Scale subscales with three groups of children

Table 2.
Means of BASC Parent Rating Scale composites with three groups of children

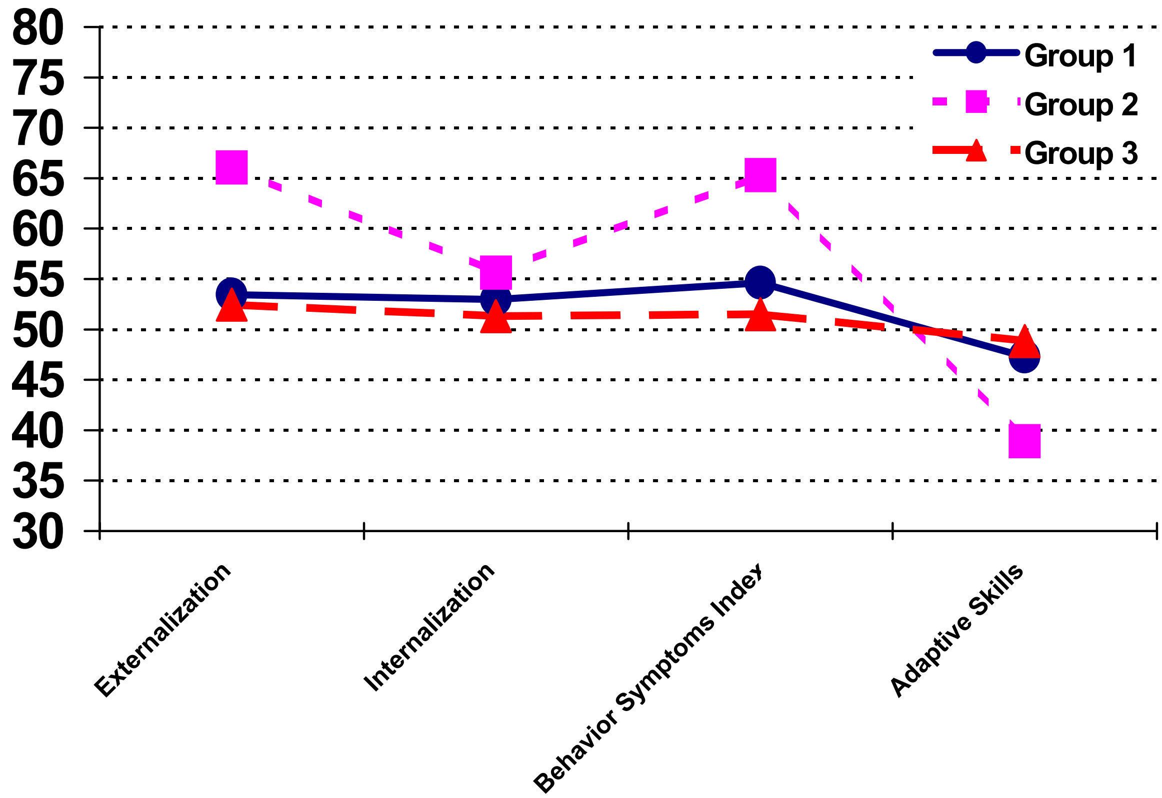
Composite Scale	Group 1		Group 2		Group 3	
	Mean	Std. Dev.	Mean	Std. Dev.	Mean	Std. Dev.
Externalization	53.43	13.55	*66.07	12.79	52.47	12.97
Internalization	52.96	11.28	55.64	11.19	51.33	11.10
Behavior Symptoms Index	54.61	16.13	*65.31	10.19	51.50	12.88
Adaptive Skills	47.30	9.41	*38.93	10.18	48.83	9.19

*Significantly different from Group 1 ($p \leq .05$)

Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture



Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture

Figure 2. Means of BASC Parent Rating Scale composite scales with three groups of children

Table 3.
Means of BASC Teacher Rating Scale subscales with three groups of children

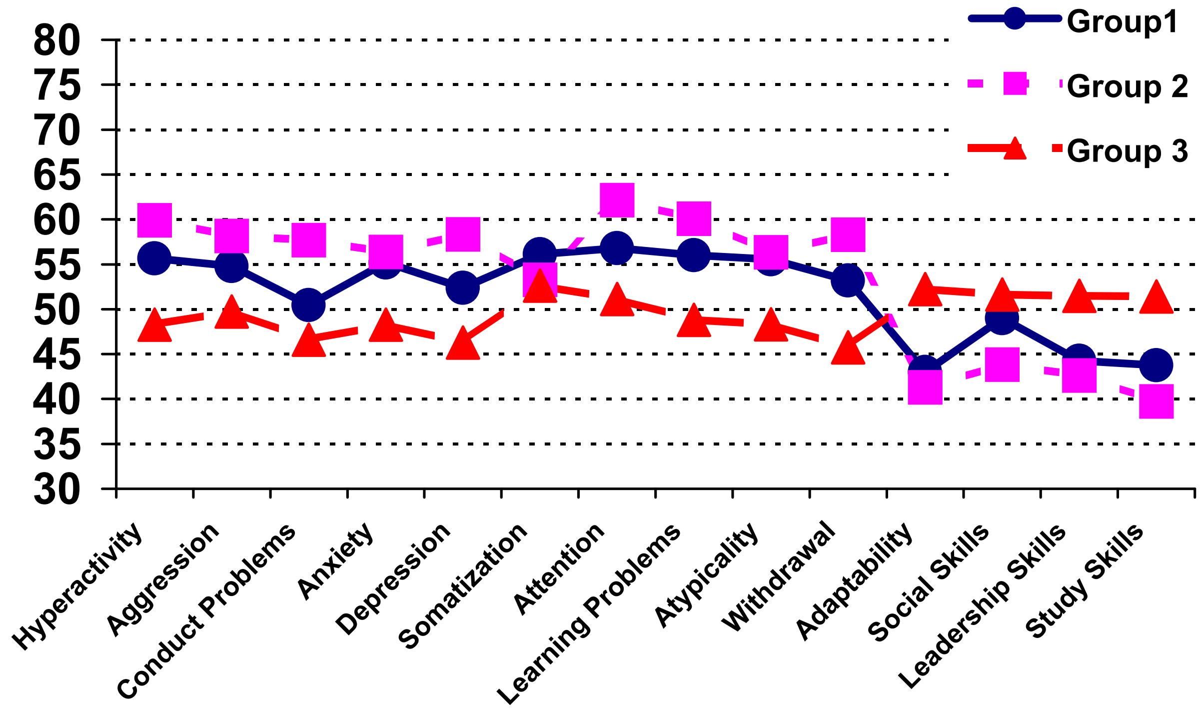
Subscale	**Group1**		**Group 2**		**Group 3**	
	Mean	Std. Dev.	Mean	Std. Dev.	Mean	Std. Dev.
Hyperactivity	55.67	7.86	59.91	10.91	*48.29	7.89
Aggression	54.81	10.86	58.15	11.84	49.76	7.15
Conduct Problems	50.44	7.57	*57.68	13.19	*46.62	2.75
Anxiety	55.19	8.46	56.38	11.77	*48.29	6.89
Depression	52.38	6.82	58.32	10.48	*46.29	5.43
Somatization	56.13	7.99	53.28	14.24	52.62	10.20
Attention	56.81	9.35	62.15	9.81	51.14	7.99
Learning Problems	56.00	10.44	60.09	10.61	*48.81	9.67
Atypicality	55.56	9.15	56.34	11.62	*48.33	4.59
Withdrawal	53.19	12.46	58.31	12.58	*45.76	6.02
Adaptability	43.00	11.14	41.31	7.70	*52.24	7.69
Social Skills	49.00	10.39	*43.84	8.92	51.62	7.87
Leadership Skills	44.25	8.87	42.60	7.08	*51.48	9.24
Study Skills	43.75	9.59	39.74	7.72	*51.43	10.71

*Significantly different from Group 1 ($p \leq .05$)

Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture



Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture

Figure 3. Means of BASC Teacher Rating Scale subscales with three groups of children

Table 4.
Means of BASC Teacher Rating Scale composites with three groups of children

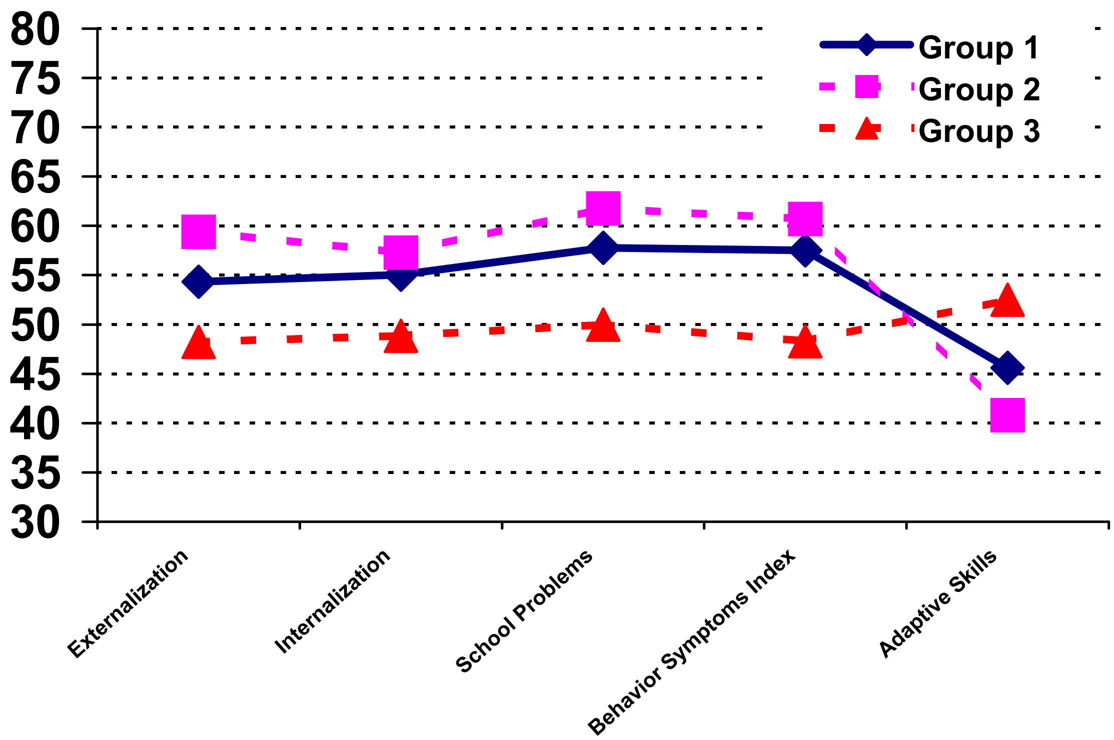
Composite Scale	**Group 1**		**Group 2**		**Group 3**	
	Mean	Std. Dev.	Mean	Std. Dev.	Mean	Std. Dev.
Externalization	54.25	8.02	59.41	11.24	*48.24	5.91
Internalization	55.31	7.46	57.29	11.73	*48.86	6.26
School Problems	56.75	9.46	61.76	9.85	*50.00	8.27
Behavior Symptoms Index	56.31	7.66	60.71	9.02	*48.31	6.26
Adaptive Skills	44.44	9.65	40.81	7.30	*52.52	9.59

*Significantly different from Group 1 ($p \leq .05$)

Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture



Group 1 = Children with a history of isolated metopic synostosis

Group 2 = Children diagnosed with ADHD

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic suture

Figure 4. Means of BASC Teacher Rating Scale composite scales with three groups of children

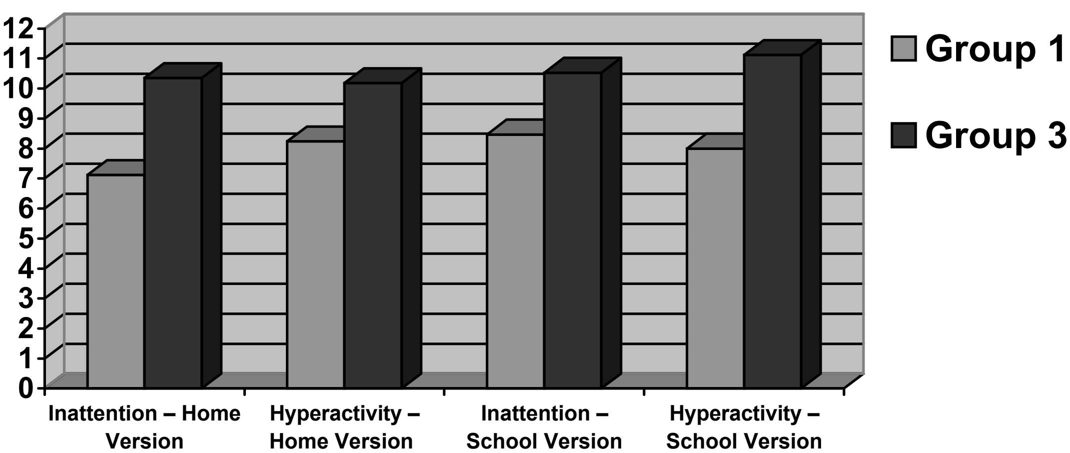
Table 5.
Mean subscale scaled scores for ADDES Home and School Versions

Subscale	**Group 1**		**Group 3**	
	Mean	Std. Dev.	Mean	Std. Dev.
Inattention – Home Version	7.12	3.20	*10.35	3.00
Hyperactivity – Home Version	8.24	3.76	10.18	2.63
Inattention – School Version	8.47	2.62	*10.53	1.97
Hyperactivity – School Version	8.00	3.61	*11.12	1.65

*Significantly different from Group 1 ($p \leq .05$)

Group 1 = Children with a history of isolated metopic synostosis

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic Suture



Group 1 = Children with a history of isolated metopic synostosis
Group 3 = Children with a history of isolated craniosynostosis not involving the metopic Suture

Figure 5. Mean subscale scaled scores for ADDES Home and School Versions

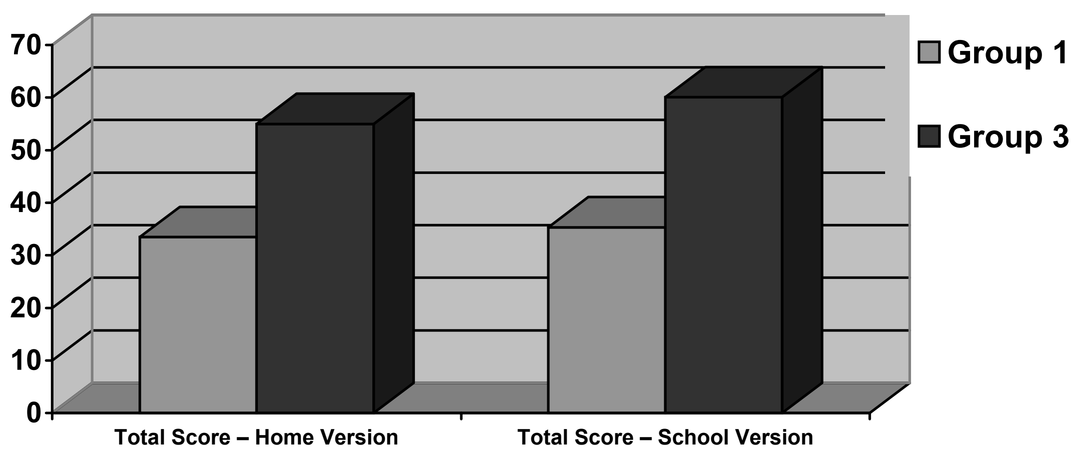
Table 6.
Mean Total Percentile Ranks for ADDES Home and School Versions

	Group 1		Group 3	
	Mean	Std. Dev.	Mean	Std. Dev.
Total Score – Home Version	33.47	23.89	*55.00	26.47
Total Score – School Version	35.35	23.98	*60.06	21.12

*Significantly different from Group 1 ($p \leq .05$)

Group 1 = Children with a history of isolated metopic synostosis

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic Suture



Group 1 = Children with a history of isolated metopic synostosis

Group 3 = Children with a history of isolated craniosynostosis not involving the metopic Suture

Figure 6. Mean Total Percentile Ranks for ADDES Home and School Versions

CHAPTER 5

DISCUSSION

The purpose of the current project was to determine if children with a history of isolated (nonsyndromic) metopic synostosis evidence a behavior pattern associated with attention deficit/ hyperactivity disorder. This goal was attempted utilizing three different groups of children. The first group of children consisted of children that had a history of isolated metopic synostosis. The second group consisted of children who had been diagnosed with attention deficit/ hyperactivity disorder and the third group consisted of children who had a history of isolated craniosynostosis not involving the metopic suture.

It was predicted that the children who had a history of isolated metopic synostosis would demonstrate a behavior pattern closely associated with ADHD. Conversely, it was predicted that children who had a history of isolated craniosynostosis not involving the metopic suture would demonstrate a dissimilar behavior pattern compared to children with a history of metopic synostosis. Behavior was measured using the BASC Parent and Teacher Rating Forms as well as the ADDES Home and School Versions.

In light of these predictions, five hypotheses were made. Hypothesis 1 stated that children with a history of isolated metopic synostosis would evidence a similar behavior pattern compared to a group of children diagnosed with ADHD as measured by the BASC Parent Rating Scale. Similarly, Hypothesis 2 stated that children with a history of metopic synostosis would evidence a similar behavior pattern compared to a group of children diagnosed with ADHD as measured by the BASC Teacher Rating Scale.

Conversely, Hypothesis 3 stated children with a history of isolated craniosynostosis not involving the metopic suture would evidence a behavior pattern that is dissimilar compared to children with a history of metopic synostosis as measured by the BASC Parent Rating Scale. Hypothesis 4 stated that children with a history of isolated craniosynostosis not involving the metopic suture would evidence a behavior pattern that is dissimilar compared to children with a history of metopic synostosis as measured by the BASC Teacher Rating Scale. Finally, Hypothesis 5 stated that children with a history of metopic synostosis would demonstrate more behaviors associated with ADHD than children with a history of isolated craniosynostosis not involving the metopic suture as measured by the ADDES Home and School versions.

The results of this study indicated that the predictions of this project were only partially realized. The results lend support to Hypothesis 2, Hypothesis 4 and Hypothesis 5. However, the results of this study repudiated Hypothesis 1 and Hypothesis 3. Consequently, the results of this study were mixed and the predictions of this study were supported only in part. However, there are some interesting trends to the results of this study.

Parent Versus Teacher Perceptions Utilizing BASC

The hypotheses (Hypothesis 1 and Hypothesis 3) that examined the perceptions of mothers regarding their child's behavior utilizing the BASC Parent Rating Scale were not supported. That is, the mothers of children with a history of metopic synostosis did not perceive that their children's behavior differed relative to the manner in which the mothers of children with a history of craniosynostosis not involving the metopic suture viewed their children's behavior. Similarly, the mothers of children with a history of metopic synostosis viewed their children in a dissimilar fashion compared to the manner in which mothers of children diagnosed with ADHD perceive their children. In sum, when utilizing the BASC Parent Rating Scale, the mothers of children with a history of metopic synostosis do not appear to view their children's behavior pattern in a manner that indicates that they suffer from ADHD. Instead, it appears that these mothers perceive their children's behavior as rather normal.

However, this trend does not hold true for the schoolteachers of children with a history of metopic synostosis. The hypotheses (Hypothesis 2 and Hypothesis 4) that examined the perceptions of schoolteachers regarding their student's behavior utilizing the BASC Teacher Rating Scale were supported. That is, the schoolteachers of children with a history of metopic synostosis did view the behavior of these children differently relative to the manner in which schoolteachers viewed children with a history of craniosynostosis not involving the metopic suture. Instead, the schoolteachers of children with a history of metopic synostosis perceived these children in a similar fashion relative to the manner in which schoolteachers view children diagnosed with ADHD when perceptions of behavior is measured using the BASC Teacher Rating Scale. Therefore, it appears that schoolteachers view the behavior patterns of children with a history of metopic synostosis in a manner that indicates that these children exhibit many of the same behaviors associated with ADHD when placed in an academic environment. This trend is significantly dissimilar to the way that schoolteachers view the behavior patterns of children with a history of craniosynostosis not involving the metopic suture.

It is unclear why mothers and schoolteachers perceive the behavior of children with a history of metopic synostosis in a different manner. That is, it is uncertain why mothers of children with a history of metopic synostosis do not appear to perceive their children as having ADHD characteristics while the schoolteachers do perceive these children as possessing ADHD characteristics. However, there are a few different possibilities that may account for these differences.

Home and Academic Environments - The first possibility centers upon the fact that the behavioral observations of mothers and teachers occur in significantly different environments. Home and school environments differ along several important dimensions. Most academic situations occur within an environment that includes a classroom full of other students as well as many other competing stimuli such as chalkboards, windows, computers etc. Furthermore, academic environments are highly structured environments that require students to focus upon certain subsets of stimuli (i.e. books, instructor) while simultaneously ignoring competing stimuli.

Children that possess characteristics of ADHD often experience difficulties maintaining attention and concentration for extended periods of time in such environments. This type of environment is significantly different from a less structured home environment in which a child is not asked to focus and concentrate upon specified stimuli for extended periods of time. Instead, children may be asked to complete shorter tasks such as “take out the garbage,” or “pick up your toys.” Tasks such as these do not require the same type of attention and concentration required to focus upon a lecture or a writing assignment within a busy classroom. Consequently, children with ADHD often experience more success completing shorter and simpler tasks within the home environment relative to their ability to complete tasks that demand attention and concentration within an academic environment full of competing stimuli.

Second, most academic classrooms consist of 15 – 25 students and one or two instructors. In an environment such as this it is often difficult if not impossible to give students individualized attention. This type of situation is significantly different from a home environment in which a child may be one of only a few siblings in the household or even the only child within the household. It becomes much easier for authority figures (i.e. mothers and fathers) to give a child individualized attention when there are only a few other children competing for attention.

This individualized attention is a great asset when asking a child to complete a complex task requiring attention and concentration such as completing homework in the evenings. Working individually with the child enables a mother or father to give their child immediate and frequent feedback or reinforcement when the child is attempting to complete a complex task that requires a significant amount of attention and concentration. As mentioned previously in this study, children who possess characteristics of ADHD attend and perform much better in an environment in which they receive immediate and frequent feedback relative to environments in which feedback is giving on a much more delayed and infrequent basis. Consequently, the teachers are likely observing behavior patterns in an environment that is much more prone to elicit behaviors associated with ADHD. Likewise, the parents of children with a history of metopic are observing the behavior patterns of these children in an environment that make the observation of behaviors associated with ADHD less probable.

Inherent Bias Among Parents - Another reason that parents and schoolteachers may perceive the behavior patterns of children with a history of metopic synostosis differently from one another stems from an inherent bias that may exist within the parents of these children. The vast majority of the children with a history of metopic synostosis utilized in this study underwent corrective craniofacial surgery as infants. This surgery is highly invasive, expensive and is accompanied by massive swelling of the cranial vault and facial region that often results in the infant's eyes swelling shut. This swelling ensues rapidly two or three hours following surgery and begins to dissipate five or six days following surgery. Furthermore, a coronal incision is made on the infant's scalp that stretches from ear to ear and is heavily stitched and bandaged following surgery. This incision leaves a lasting scar under the child's hair.

The infants who are subjected to this surgery appear to go unaffected psychologically or emotionally. However, the entirety of this experience can be emotionally distressing for the parents of these young children. Consequently, parents may be emotionally invested to perceive their child as being normal. That is, perceiving their child as being normal may confirm their belief that the craniofacial surgery was in the best interest of the child and may relieve any feelings of guilt associated with subjecting their young infant to an invasive surgical procedure.

On the other hand, many of the schoolteachers of these children may be completing unaware of the child's medical history. Additionally, in the event that some of the schoolteachers were aware of the child's medical history, presumably none of them experienced the vicarious distress of watching the child undergo craniofacial surgery and the accompanying recovery period. As a result, the schoolteachers are far less likely to possess an inherent bias to perceive the behavior patterns of these children as normal.

Parent and Teacher Perceptions Utilizing ADDES

Although the results of the present study were mixed when utilizing the BASC Parent and Teacher Rating Scale, the results were more definitive when utilizing the ADDES Home and School Versions. Hypothesis 5 predicted that children who have a history a metopic synostosis would evidence more behaviors associated with ADHD than children who have a history of isolated craniosynostosis not involving the metopic suture. The Total scores of both the ADDES Home and School versions, the Inattentiveness and Hyperactivity subscales of the ADDES School Version and the Inattentiveness subscale of the ADDES Home Version supported this prediction. The only test result that did not support Hypothesis 5 was the one that indicated no significant difference existed between the two groups of children on the Hyperactivity subscale of the ADDES Home Version. Instead, this test result supported the trend that mothers of children with a history of metopic synostosis do not view their children differently relative to mothers of children with a history of craniosynostosis not involving the metopic suture.

It is interesting to note that while hypotheses (Hypothesis 1 and 3) examining maternal perceptions of behavior patterns utilizing the BASC Parent Rating Forms were not supported, the results of Hypothesis 5 generally did support predicted maternal perceptions. It is unclear why the mothers of children with a history of metopic synostosis would rate the behaviors of these children in an apparently discrepant manner from one behavior-rating questionnaire to another. However, there are a couple of explanations that may account for these results.

Focus of the ADDES Home Version

The focus of the BASC Parent Rating Scale and the ADDES Home Version are different from one another. Simply speaking, the focus of the BASC Parent Rating Scale is broad while the focus of the ADDES Home Version is narrow. The BASC Parent Rating Scale was developed in order to allow for a more global assessment of childhood behavior patterns. This allows researchers and clinicians to assess for the presence of any number of maladaptive and adaptive behavior patterns utilizing a single instrument. The maladaptive behavior patterns may include depression, anxiety, somatization etc, in addition to the hyperactivity and inattentiveness associated with ADHD. On the other hand, the ADDES Home Version was developed to specifically assess for the presence of behaviors associated with ADHD as outlined by the DSM-IV. Consequently, this measure is much specific and narrowly defined. Additionally, given the fact that the items from the ADDES Home Version stem directly from DSM-IV diagnostic criteria for ADHD, it may provide a more valid assessment of this disorder than the BASC PRS, which does not directly assess the DSM-IV diagnostic criteria for ADHD.

The difference in focus between these two measures may partially account for the discrepant results obtained in this study. Given the fact that the focus of the two measures is different from one another may elicit differing response patterns on the part of the raters. It is possible that the mothers of children with a history of metopic synostosis may respond to questions concerning ADHD behaviors in a sensitive manner when completing the ADDES Home Version relative to the BASC Parent Rating Scale because it is more narrowly focus. Conversely, these same raters may not respond as sensitively to questions concerning ADHD when completing the BASC Parent Rating Scale because items addressing other behaviors may "cloud" items addressing ADHD behaviors. In essence, the global focus of the BASC Parent Rating Scale may make it less sensitive to behaviors associated with ADHD relative to the ADDES Home Version.

Behavioral Anchors of the ADDES and BASC

In addition to the focus of the BASC and the ADDES being different from one another, these two measures also utilize different behavioral anchors that may partially account for the discrepant results. All of the BASC rating forms measure behavior on a 4-point scale of frequency from (1) never to (4) almost always. The ADDES forms measure behavior on a 5-point scale of frequency from (0) Does Not Engage In The Behavior; to (4) Engages In The Behavior One To Several Times Per Hour. The behavioral anchors used with the ADDES forms offer parents and teachers more behavioral descriptors, which may enable them to rate the child's more accurately. Again, the behavioral descriptors that are used with the ADDES forms are possible because this measure focuses upon a narrower domain of behavior relative to the BASC instruments, which have a more global focus.

Nature of Comparisons

The apparently discrepant results obtained in this examination may also partially result from the nature of the comparisons made in this study. The BASC Parent Rating Scale compared behavior patterns of children with a history of metopic synostosis with children diagnosed with ADHD as well as children with a history of craniosynostosis not involving the metopic suture. However, comparisons were only made between children with a history of metopic synostosis and children with a history of craniosynostosis not involving the metopic suture when utilizing the ADDES Home and School Versions.

Consequently, it can generally be said that the results of this study support the hypothesis that children with a history of metopic synostosis exhibit more behaviors associated with ADHD relative to children with a history of craniosynostosis not involving the metopic suture as measured by the ADDES Home Version. However, it cannot be said that children with a history of metopic synostosis exhibit as many behaviors associated with ADHD as children diagnosed with ADHD. This type of comparison was not made using the ADDES Home Version. It may be that children with a history of metopic synostosis differ significantly from children diagnosed with ADHD as measured by the ADDES Home Version. That is, it is possible that children with a history of metopic synostosis evidence fewer behaviors associated with ADHD than children diagnosed with ADHD but more behaviors associated with ADHD than children with a history of craniosynostosis not involving the metopic suture as measured by the ADDES Home Version. A finding such as this would lend support to the trend that mothers of children with a history of metopic synostosis rate their child's behavior patterns differently relative to the way mothers of children diagnosed with ADHD rate their children's behavior.

Strength of Experimental Design

The apparently discrepant results of this examination also may have partially resulted from the strength of the experimental design. Comparisons of behavior patterns between children with a history of metopic synostosis and children with a history of craniosynostosis not involving the metopic suture using the ADDES Home and School Versions were made utilizing subjects that were matched in terms of age and sex. This made it possible to yoke each of the subjects, which increased the strength of the experimental design and statistical power. Consequently, the ability to match subjects in terms of age and sex increased the likelihood that a statistical difference would be found between the two groups should any differences exist.

On the other hand, the comparisons between the three groups of children when using the BASC Parent and Teacher Rating Scales were not made utilizing subjects that had been matched in terms of age and sex. This was due to the fact that the data for the group of children diagnosed with ADHD was obtained from a normative sample used during the development of the BASC Parent and Teacher Rating Scales. Given the fact that the occurrence of isolated craniosynostosis is very low, it would be extremely difficult to age and sex match children with a history of craniosynostosis with the children in the ADHD reference group. As a result, the strength of this experimental design is not as strong and offers less statistical power than the design utilized when comparing ADDES Home and School Version results. This weakness may partially account for lack of dissimilarity found between the children with a history of metopic synostosis and the children with a history of craniosynostosis not involving the metopic suture when utilizing the BASC Parent Rating Scale.

Trends Common to BASC and ADDES Comparisons

Despite the fact that the present examination produced mixed results, there were some common trends that are very interesting. First, all of the hypotheses (Hypothesis 2, Hypothesis 4 and half of Hypothesis 5) that focused upon the perceptions of schoolteachers in regards to the behavior patterns among children with a history of metopic synostosis were supported. The results of these hypotheses suggest that the schoolteachers of children with a history of metopic synostosis perceive these children in a manner that indicates these children exhibit more behaviors associated with ADHD and are more similar to a group of children diagnosed with ADHD than a control group.

This trend is especially noteworthy given the fact that parents may be hesitant to perceive children with a history of metopic synostosis as being abnormal for reasons previously discussed. These reasons suggest that the results obtained from the schoolteachers may offer a more valid and accurate account of behavior patterns than the results obtained from the mothers of these children. Using this presupposition, the current examination offers intriguing support for the notion that children with a history of metopic synostosis exhibit more ADHD characteristics than children with a history of craniosynostosis not involving the metopic suture.

A second noteworthy trend also concerns the manner in which mothers of children with a history of metopic synostosis perceive their children's behavior relative to the manner in which the schoolteachers perceive the behavior of the same children. As already discussed, significant differences were found when comparing the two groups of children with craniosynostosis on the Total scores of both the ADDES Home and School versions, the Inattentiveness and Hyperactivity subscales of the ADDES School Version and the Inattentiveness subscale of the ADDES Home Version. Above and beyond these differences, it is interesting to note that the effect sizes were generally larger for comparisons that were made utilizing the ADDES School Version relative to the effect sizes found for ADDES Home Version.

First, a larger effect size was found for the Hyperactivity subscale of the ADDES School Version (group mean difference of 3.12) relative to the effect size found for the

Hyperactivity subscale of the ADDES Home Version (group mean difference of 1.94). Additionally, the effect size for the Total Score of ADDES School Version (group mean difference of 24.71) was larger than the effect size obtained for the Total Score of the ADDES Home Version (group mean difference of 21.53). These trends again supports the notion that the schoolteachers of children with a history of metopic synostosis may be more sensitive to behavior patterns associated with ADHD than are the mothers of the same children.

Weaknesses of Examination

Despite the interesting findings offered by the current examination, there are some weaknesses that limit the conclusions that can be drawn from this study. First, all of the findings in this study are based solely upon the responses that mothers and schoolteachers gave on two different standardized behavior-rating questionnaires. Behavioral assessment was conducted in this manner because most of the children used in this study lived great distances from the site of the investigation and were unable to travel to the site in order to be evaluated with more direct measures. Consequently, all behavioral assessment needed to be conducted through the postal service.

It is true that standardized behavior-rating questionnaires offer several advantages. These advantages include the ability to gather behavioral information from an observer that has had many interactions with the child in a wide variety of settings. Standardized behavior-rating questionnaires also offer to the ability to quantify and assess the qualitative aspects of behavior.

However, the results of behavior-rating questionnaires are based upon the subjective perceptions of the rater, which may or may not accurately reflect actual behavior. Consequently, firm conclusions concerning children with a history of metopic synostosis can not be reached until a study that utilizes direct behavioral observation is conducted. Direct behavioral observations would offer an avenue for evaluating the accuracy of the responses offered by the raters of behavior-rating questionnaires.

It would also be advantageous to utilize neuropsychological instruments that are designed to measure prefrontal cortex functioning in future studies given the presupposition that behavior patterns associated with ADHD stem from prefrontal cortex dysfunction. Utilizing neuropsychological instruments such this would afford researchers two advantages. First, it would enable researchers to compare the performance of children with a history of metopic synostosis with normal populations as well clinical populations suffering from deficits with attention and executive function. Second, it would afford researchers the opportunity to directly observe behavior under standardized conditions.

The present study was also weakened by the fact that the age range of both groups of children with a history of craniosynostosis was 4 years through 12 years of age. This age range is not a limitation when using the ADDES Home and School Versions. However, this age range is a significant limitation when utilizing the BASC Parent and Teacher Rating Scales given the fact that there is separate rating forms for children and preschoolers. Consequently, the mothers and teachers of children 4 –5 years of age rated subjects on different BASC forms than the mothers and teachers of children 6 – 12 years of age. These differing forms contain different individual items that reflect developmental differences between preschoolers and children 6 – 12 years of age. It would have been ideal to limit the subjects in the present study to one of the age ranges (4-5 years or 6-12 years). However, given the low incidence of craniosynostosis it was impossible to limit the study to one of these age ranges. To do so would have significantly restricted the total sample size. Consequently, it must be presupposed that analogous scales on the two forms of the BASC measure the same behavioral constructs despite containing different individual items.

Another limitation of the current study is that it failed to control for several factors associated with craniosynostosis including age of onset, age of craniofacial surgery, severity and reoccurrence of disorder as well as the amount of time between the onset and correction of the disorder. It is unclear what neuropsychological and neuro-behavioral effects may result from these different factors associated with craniosynostosis. The difficulty with a study designed to control for some or all of these factors is that craniosysnostosis as a whole is a disorder that occurs in 0.4/1,000 births (Hunter & Rudd, 1976), while metopic synostosis accounts only 3-4% of all synostoses (Hunter & Rudd, 1976). Consequently, it would take a commendable undertaking to recruit and obtain the appropriate patients to control for some or all of these factors.

It is interesting to note, however, that a recent study conducted by Kapp-Simon (1998) found that rates of mental retardation and learning disabilities were not influenced by surgical status of children with craniosynostosis. That is, children who did not have corrective surgery were found to be no likely to suffer from mental retardation and learning disabilities than children who did have corrective surgery. Furthermore, the hypothesis that early surgical intervention of the craniosynostosis would improve mental functioning during childhood was not upheld in this study.

A similar limitation of the current study is that it failed to control for certain psychosocial factors. Most notably, this study did not control for the socioeconomic status of the children utilized in this examination. It has been found that children differing in terms of socioeconomic status frequently differ in terms of behavior patterns as measured by standardized behavior-rating questionnaires. Therefore, it is often sound theoretical practice to control for this factor when comparing the behavior patterns of two groups of children. Once again, however, a study controlling for this factor would be difficult to achieve given the infrequent incidence of craniosynostosis.

A final limitation of the present study is that is was limited by small sample sizes. The sample sizes of the groups containing children with a history of metopic synostosis and children with a history of craniosynostosis not involving the metopic suture was limited due to the fact that craniosynostosis is a rare disorder and it is difficult to locate patients several to many years following treatment. A larger multi-site study would need to be instituted in order to obtain larger sample sizes.

The sample sizes for the ADHD clinical samples were also relatively small. However, since the completion of this examination a considerable amount of research has been conducted examining the clinical utility of the BASC Scales in the assessment of children diagnosed with ADHD. Consequently, the sample sizes of ADHD clinical samples are much larger at the present time. It would be interesting and informative to conduct the present examination again using these larger BASC ADHD clinical samples.

The larger sample sizes would allow for more accurate and refined comparisons between children diagnosed with ADHD and children with a history of craniosynostosis. For example, the ADHD clinical sample used in the present study was a heterogeneous group of children. That is, it contained children who were both ADHD, Predominately Inattentive-Type and children who were ADHD, Predominately Hyperactive-Type. The larger sample sizes now available through American Guidance Service, Inc. (AGS) would provide the ability to obtain more homogenous ADHD clinical samples. It would then be possible to compare children with a history metopic synostosis with children diagnosed with ADHD, Predominately Inattentive-Type and children diagnosed with ADHD, Predominately Hyperactive-Type.

The larger ADHD sample sizes also make it possible to delineate which subscales and which composite scales are the most accurate for distinguishing ADHD children from non-ADHD children. It would then be possible to make group comparisons utilizing only the most sensitive scales while eliminating less useful scales. Minimizing the number of scales to only the most sensitize scales would allow for more sensitive and accurate group comparisons.

Strengths of Examination

The primary strength of the present examination is that it significantly contributes to the understanding of the long-term behavioral consequences associated with isolated metopic synostosis. Sidoti, Marsh, Marty-Grames and Noetzel (1996) conducted the only other study that has attempted to examine the behavioral consequences of metopic synostosis. Again, this study was purely descriptive in nature and included 32 cases of children who had a history of metopic synostosis. Utilizing a very brief questionnaire that was filled-out by the parents of these children, four of these patients were identified as having "attention deficit/hyperactivity disorder," four patients were reported as being "mentally retarded" and three were described as having "speech and language delays." However, this study did not utilize standardized measures or a control group. Consequently, it is very difficult to ascertain the accuracy of the descriptions or diagnoses obtained in this study or their significance relative to a control group.

The present examination, however, utilized four different standardized behavior-rating questionnaires that were completed by two independent raters. This examination also included two different control groups, which also enhanced the ability to draw more firm conclusions regarding the behavior patterns of children with a history of metopic synostosis. These improvements provide valuable information concerning the long-term behavioral consequences associated with metopic synostosis. Additionally, the results of this examination provide further support for the hypothesis that children with a history of metopic synostosis are at risk for the development of a behavior pattern associated with ADHD.

CHAPTER 6

CONCLUSIONS

The purpose of this project was to examine the behavior patterns among children with a history of metopic synostosis. It was hypothesized that children with a history of metopic synostosis would exhibit many of the same behavioral patterns associated with ADHD. It was also hypothesized that children with a history of simple synostosis not involving the metopic suture would not evidence this type of behavioral pattern.

It was found using the Home and School versions of the Attention Deficit Disorders Evaluation Scale (ADDES) that children with a history of metopic synostosis demonstrate significantly more behavioral disturbances than children with a history of simple craniosynostosis not involving the metopic suture. Using the BASC Teacher Rating Form it was found that children with a history of metopic synostosis have a behavior pattern similar to children diagnosed with ADHD and a dissimilar behavior pattern compared to children who have a history of craniosynostosis not involving the metopic suture. Using the BASC Parent Rating Form it was found that children with a history of metopic synostosis have a behavior pattern dissimilar to children diagnosed with ADHD and a dissimilar behavior pattern compared to children who have a history of craniosynostosis not involving the metopic suture.

These results taken together lend modest support to the notion that children with a history of metopic synostosis are at risk for developing ADHD-type behavior patterns as they mature. While this examination adds to the existing literature that addresses this issue, it falls significantly short of offering definitive evidence for concluding that children with a history of metopic synostosis are at increased risk for developing ADHD. However, it adds considerably to a body of literature that has examined this issue and offers further evidence that more research needs to be conducted in this area. More research such as this needs to be conducted in order for health care professionals to provide better answers concerning the long-term impact of craniosynostosis to the families of children diagnosed with this disorder. In order to obtain definitive results, researchers in this area need to begin to collaborate on larger, multi-center studies in order to obtain larger sample sizes and control for medical and psychosocial factors that may influence cognitive development.

Appendix A

Letter to Parents

Brad Kuper, M. A.
Behavioral Medicine for Children
Medical City Hospital Dallas
7777 Forest Lane
Dallas, TX
(972) 566-6724

Dear Sir or Maam:

The Cranio-Facial Institute and the Department of Behavioral Medicine at Medical City Hospital Dallas are currently undertaking an investigation that is designed to examine the behavior patterns of children who have been treated for craniosynotsosis. Consequently, we are asking you to take a few minutes and rate your child's behavior using the behavior rating forms enclosed with this packet of materials. The behavior ratings forms include the the Behavior Assessment System for Children Parent Rating form and the Attention Deficit Disorders Evaluation Scale - Parent Version. In addition, we would also like you to complete the Developmental History questionaire that we have included. In the past it has been found that mothers and fathers often rate their child's behavio very differently. Therefore, we prefer that the mothers of the participating families complete these behavior rating forms in order to control for this factor.

We also are asking that you allow your child's primary school teacher to complete two behavior rating forms in order to obtain a more complete picture of your child's behavior at home and at school. The Behavior Assessment System for Children Teacher Rating form and the Attention Deficit Disorders Evaluation Scale - Teacher Version are enclosed in the envelope entitled "Teacher Rating Forms." These two rating forms are very similar to the rating forms that you will be completing. We have also enclosed a letter and a "Teacher Informed Consent Form" that will describe the nature and the purpose of this project to your child's school teacher.

Before you agree to partcipate in this study please read the "Parent Informed Consent Form" that is enclosed in the packet of materials. If you agree to participate please sign and date these forms before completing the behavior rating forms and developmental history. Once you have completed these forms please return them, along with the Parent Informed Consent Form" in the self addressed envelope that has been provided. In addition, please deliver the envelope entitled "Teacher Rating Forms" to your child's primary school teacher. Your child's teacher will complete the questionaires that are included in this envelope and return them in a self-addressed envelope that is provided. Please complete these materials within a two week period.

Thank you for taking the time to review and complete these materials. The information that you will be providing the Cranio-Facial Institute and the department of Behavioral Medicine for Children will enable everyone associated with the Cranio-Facial Insitute to more adquately address the needs of families and children that have been treated for craniosynostosis. If you have any questions regarding the nature or purpose of this project please feel free to call us at (972) 566-6724.

Sincerely,

Behavioral Medicine for Children
Medical City Hospital Dallas

Appendix B

Letter to Teachers

Brad Kuper, M. A.
Behavioral Medicine for Children
Medical City Hospital Dallas
7777 Forest Lane
Dallas, TX
(972) 566-6724

Dear Sir or Maam:

The Cranio-Facial Institute and the Department of Behavioral Medicine at Medical City Hospital Dallas are currently undertaking an investigation that is designed to examine the behavior patterns of the children who have visited our clinic. Consequently, we are asking you to take a few minutes and rate your student's behavior using the behavior rating forms enclosed with this packet of materials. The Behavior Assessment System for Children Teacher Rating form and the Attention Deficit Disorders Evaluation Scale - Teacher Version are enclosed in this envelope.

Before you agree to partcipate in this study please read the "Teacher Informed Consent Form" that is enclosed in the packet of materials. If you agree to participate please sign and date these forms before completing the behavior rating forms. Once you have completed these forms please return them, along with the "Teacher Informed Consent Form" in the self addressed envelope that has been provided. Please complete these materials and return them within a two week period.

Thank you for taking the time to review and complete these materials. The information that you will be providing the Cranio-Facial Institute and the department of Behavioral Medicine for Children will enable everyone associated with the Cranio-Facial Insitute to more adquately address the needs of families and children that have been treated for craniosynostosis. If you have any questions regarding the nature or purpose of this project please feel free to call us at (972) 566-6724.

Sincerely,

Appendix C
Parent Informed Consent Form

Behavioral Medicine for Children
Medical City Hospital Dallas
7777 Forest Lane
Dallas, TX
(972) 566-6724

Contact Persons:
Susan Porter-Levy, Psy.D.
Jerry McGill, Ph. D.
Brad Kuper, M. A.

Description: The purpose of the present study is to examine the impact that craniosynostosis has upon the behavior patterns of children. In order to accomplish this goal you are being asked to rate your child's behavior using two different ratings scales and a developmental questionnaire. The first rating scale is called the Behavior Assessment System for Children parent rating form (BASC; Reynolds and Kamphaus, 1992). This form measures behavior on a 4-point scale of frequency from (1) Never to (4) Almost Always and assesses the child's adaptive and problem behaviors at home as well as in social situations. It takes approximately 20 minutes to complete. The second rating form is called the Attention Deficit Disorders Evaluation Scale - Parent Version (ADDES; McCarney, 1995). This form measures behavior on a 5-point scale of frequency from (0) Does Not Engage In The Behavior; to (4) Engages In The Behavior One To Several Times Per Hour. It takes approximately 10 minutes to complete. Finally, the BASC developmental history questionnaire should take approximately 15 minutes to complete.

Be sure to complete all of the items and all of the demographic information for each of the questionnaires. After you have completed all of the materials please return them, including this informed consent form, in the self-addressed envelope that has been provided. Please return these materials within a two-week period.

We also are asking to allow your child's primary school teacher the opportunity to answer questions regarding your child's behavior school. However, you child's teacher will not be notified that your child has a history of craniosynostosis. In addition to the parent rating forms just described we have also included teacher rating forms within the envelope entitled "Teacher Rating Forms." This envelope contains four items. First, it contains an informed consent form very similar to the one you are currently reading that describes the nature and the purpose of the current project. Second, it contains the Behavior Assessment System for Children teacher rating form which is similar to the Behavior Assessment System for Children parent rating form that you are being asked to complete. Third, it contains the Attention Deficit Disorders Evaluation Scale - Teacher Version form which is very similar to the Attention Deficit Disorders Evaluation Scale -

Parent Version form that you are being asked to complete. Fourth, it contains a self-addressed envelope. Please instruct your child's teacher to complete these materials within a two week period.

Confidentiality: The tests used during this procedure will be scored only by one of the primary investigators listed above. Furthermore, only group scores and averages will be used during analysis. At no time during the analysis will individual scores be used or examined. In addition each participant in the study will be given a number that will be used to identify scores and to ensure confidentiality. As a result, names will not be used and individual scores can not be linked to individuals' names. People with access to the raw data will be the contact persons listed above. Data will be kept in a locked file cabinet.

Risks and Benefits: The benefits of this project include aiding researchers in determining the effects that craniosynostosis has upon behavior patterns. This procedure is largely risk free, with the exception that parents or teachers may become sensitized to any unusual behavior patterns on the part of their child or student. If this occurs, you are encouraged to contact the researchers at the number provided during regular business hours.

Right to Withdraw: You may withdraw from this study at any time without penalty.

Voluntary Consent Statement: I have read this document and have been given the opportunity to call one the primary investigators at the number provided and ask questions regarding this study. Any questions I might have had have been answered to my satisfaction. I understand that any further questions I should have be directed to one of the contact persons listed above.

My signature below means that I understand the nature of the study and agree to participate. My signature below also means that I agree to let my child's teacher participate as described above.

Child's Name

Name of Parent or Legal Guardian

Signature of Parent or Legal Guardian

Date

APPENDIX D

Teacher Informed Consent Form

Behavioral Medicine for Children
Medical City Hospital Dallas
7777 Forest Lane
Dallas, TX
(972) 566-6724

Contact Persons:
Susan Porter-Levy, Psy.D.
Jerry McGill, Ph. D.
Brad Kuper, M. A.

Description: The purpose of the present study is to examine the behavior patterns of children that have visited our department at Medical City Hospital Dallas. In order to acccomplish this goal you are being asked to rate your student's behavior using two different ratings scales. The nature and purpose of this project have already been described to the parents of this student and they have agreed to allow you to participate. The first rating scale is called the Behavior Assessment System for Children teacher rating form (BASC; Reynolds and Kamphaus, 1992). This form measures behavior on a 4-point scale of frequency from (1) Never to (4) Almost Always and assesses the child's adaptive and problem behaviors at home as well as in social situations. It takes approximately 20 minutes to complete. The second rating form is called the Attention Deficit Disorders Evaluation Scale - Teacher Version (ADDES; McCarney, 1995). This form measures behavior on a 5-point scale of frequency from (0) Does Not Engage In The Behavior; to (4) Engages In The Behavior One To Several Times Per Hour. It takes approximately 10 minutes to complete.

Be sure to complete all of the items and all of the demographic information for each of the questionnaires. After you have completed all of the materials please return them, including this informed consent form, in the self-addressed envelope that has been provided. Please return these materials within a two-week period.

Confidentiality: The tests used during this procedure will be scored only by one of the primary investigators listed above. Furthermore, only group scores and averages will be used during analysis. At no time during the analysis will individual scores be used or examined. In addition each participant in the study will be given a number that will be used to identify scores and to ensure confidentiality. As a result, names will not be used and individual scores can not be linked to individuals' names. People with access to the raw data will be the contact persons listed above. Data will be kept in a locked file cabinet.

Risks and Benefits: The benefits of this project include aiding researchers in examining the behavior patterns of the children that have visited our department. This procedure is largely risk free, with the exception that parents or teachers may become sensitized to any unusual behavior patterns on the part of their child or student. If this occurs, you are encouraged to contact the researchers at the number provided during regular business hours.

Right to Withdraw: You may withdraw from this study at any time without penalty.

Voluntary Consent Statement: I have read this document and have been given the opportunity to call one the primary investigators at the number provided and ask questions regarding this study. Any questions I might have had have been answered to my satisfaction. I understand that any further questions I should have be directed to one of the contact persons listed above.

My signature below means that I understand the nature of the study and agree to participate.

Child's Name

Name of Child's Teacher

Signature of Child's Teacher

Date

This project has been reviewed and approved by the University of North Texas Commitee for the Protection of Human Subjects (940) 565-3940 and by the Institutional Review Board at Columbia's Medical City Hospital Dallas (972) 566-6060.

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