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The Effects of Color Preference on Attention Span in Individuals with Angelman syndrome

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This paper meets the requirements for a senior honors thesis in the department of psychology at Carroll College.

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The Effects of Color Preference on Attention Span in Individuals with Angelman syndrome

Susan Goffinet

Carroll College, 1997
There are so many people to thank, I really don’t even know where to start. This was one of the most difficult projects I have ever had to complete in all of my twenty-two years, and it would not have been possible without the support, dedication, time, commitment, energy and vast knowledge of some very important people.

First, I’d like to thank Lacey, my very special friend and “Angel,” as so many refer to their Angelman children. She was my first exposure to the disorder, and I have been fascinated by it ever since. There is no doubt how much I love her, and all of the special children with whom I work. To all of the people involved in this project, the children, parents and scientists. Your time and knowledge made this paper.

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To Robb Jones and Toni Person, the two of you have been so much more to me than “supervisors.” You are dear friends, without whom, I don’t know where I would be or what I would be doing. Thank you, Robb for threatening to fire me if I didn’t go back to school, and for listening and offering the wisest advice when I called crying about my future, or possible lack thereof. Toni, thank you for all the advice and concern over the last four years, and for all the talks about school, work, boys…. Thanks for always pulling the right strings, for Disneyland memories and for the most comfortable spare bed in the state of Idaho.

I also must thank all of my friends at work. You know who you are. I have always felt welcome, and I know I have many friends to turn to for support, advice and answers to life’s more difficult questions.

To all of my friends, and to my fellow students at Carroll. You’ve seen me in good moods and in bad moods, at my best and at my worst, and you’ve stuck by me through thick and thin. I hope you all know I would do anything for you, which still would not be enough to repay you for what you have given me. There are far too many names to name, but you know who you are.

Most importantly, I need to share my unconditional love and devotion to my parents. I have never doubted your love and dedication to me and my success in this world, as a member of society, and as a human being. You never even gave me a chance to doubt it. I have always felt safe and taken care of. There is no better feeling in the world, and I hope you know how much I appreciate you for just being “Mom” and “Dad.” I love you. And to my brother, Michael, even though we pick on each other constantly, you know I love you, and I am proud of you and all you are doing with your life. Because of you, I always have someone better to look up to.
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Abstract

Angelman syndrome is a genetic disorder involving a deletion on the maternal chromosome 15 characterized by a variety of physical, mental and behavioral problems. This paper addressed the physical problem of hypopigmentation as it affects the eyes and vision, as well as the behavioral problems of hyperactivity and a short attention span. The hypothesis is that a preference for the color yellow, which may be connected with hypopigmentation, can be used in training programs to increase attention span towards certain tasks. Five individuals with Angelman syndrome were observed to discern a color preference or a tendency to choose one color significantly more often than others. Once this was established, this information was used to test the length of attention span towards activities using the preferred color compared to the other colors. Results showed that each child did seem to prefer one color over others, but each child preferred a different color, so the color yellow was not significant. Attention span was not influenced by color at all. The importance of this paper is the idea that attention span may be increased through sensory integration types of techniques rather than through the use of drugs or other methods. This is important information for families, teachers, therapists, and care-givers who work with children who have Angelman syndrome and struggle daily, finding themselves discouraged with training programs that are inhibited by the child’s lack of attention to the task. Additional methods such as those described in this paper can be attempted to help facilitate training. Questions such as those raised in this paper offer encouragement to those working with children with Angelman syndrome to not give up on training, but to instead reevaluate the reasons behind failure and to attempt new strategies that may be better suited to the child and the situation.
The Effects of Color Preference on Attention Span in Individuals with Angelman syndrome

Reason Behind This Research Project

I began my research on Angelman syndrome after an experience working as a care-provider for a child with Angelman syndrome. I was exposed to a very limited amount of information on the disorder, and due to a very loving relationship with this child, I wanted to learn more about her condition.

The idea behind this research project came from those experiences I have had working with children who have Angelman syndrome. As a caregiver, I have been responsible for running training programs aimed at personal care skills and communication skills. Dealing with a child that has Angelman syndrome, while rewarding, can be frustrating at times because of the short attention span. Many times, the child is distracted so quickly and so often that programs seem to lose their value. It is difficult for the child to learn from the programs or to accomplish his or her goals when distractions keep him or her from finishing the task at hand.

Through conversations and correspondence with parents and care-providers; I learned that many of their children preferred yellow toys and objects to those of other colors, and thus became curious as to the possible link between ocular albinism in Angelman syndrome patients and an apparent color preference for yellow.

The objective of this project was to determine if a color preference can improve attention-span, the results from which could be used to address the behavioral problem of hyperactivity. These behavioral problems were particularly important to care-providers in that they tend to interfere with specific training attempts and protocol. Many parents believe hyperactivity in Angelman syndrome is different from those children without Angelman syndrome (Angelman Research Group, 1990). "The hyperactivity in children with Angelman
syndrome seems to be 'hypermotoric' and generally related to intense curiosity. However, the short attention span seems to keep the child 'on the go,' (Angelman Research Group, 1990). The hypothesis is that a demonstrated color preference, especially toward the color yellow as previously described, can increase attention span towards specific tasks.

In addition to the behavioral problems described above, three physical problems to be described later—ocular hypopigmentation, hypersensitivity and a sleep disorder—which may contribute in part to hyperactivity, are also important considerations when analyzing the results of this project.
What is Angelman Syndrome

Angelman syndrome is a neurological disorder caused by a chromosomal deletion. It was first diagnosed by Dr. Harry Angelman, a pediatric physician from Great Britain. In 1965, Dr. Angelman noted several unique traits shared by three children he was treating at the time. These traits included a movement and balance disorder characterized by a stiff and jerky gait; a happy and smiling demeanor with excessive laughter; a seizure disorder; and a flattened back of the head. He originally referred to these children as "puppet children," as their gait reminded him of a marionette, and the smile resembled that of a boy in a painting entitled "Boy With A Puppet." The following is Dr. Angelman's recent account of the story:

The history of medicine is full of interesting stories about the discovery of illnesses. The saga of Angelman syndrome is one such story. It was purely by chance that nearly thirty years ago three handicapped children were admitted at various times to my children's ward in England. They had a variety of disabilities and although at first sight they seemed to be suffering from different conditions I felt that there was a common cause for their illness. The diagnosis was purely a clinical one because in spite of technical investigations which today are more refined I was unable to establish scientific proof that the three children all had the same handicap. In view of this, I hesitated to write about them in the medical journals. However, when on a holiday in Italy I happened to see an oil painting in the Castelvecchio museum in Verona called "A Boy with a Puppet." The boy's laughing face and the fact that my patients exhibited jerky movements gave me the idea of writing an article about the three children with the title of "Puppet Children." It was not a name that pleased all parents but it served as a means of combining the three little patients into a single group. Later the name was changed to Angelman syndrome. This article was published in 1965 and after some initial interest lay almost forgotten until the early eighties. In fact many doctors denied that such a condition existed. In the past ten years American and English doctors have placed the syndrome on a firm footing with the ability to establish the diagnosis beyond doubt... (NASF, 1996)

According to the Angelman syndrome information packet for parents, the diagnosis is most often suggested clinically by a pediatrician who is familiar with the syndrome. It is followed by a diagnosis by a neurologist or a geneticist (National Angelman Syndrome Foundation—NASF, 1996). Diagnosis usually takes place between the ages of three and seven
years when the characteristic behaviors and facial features of Angelman syndrome become most evident (NASF, 1996).

Clinically, Angelman syndrome patients carry a variety of distinctive traits. These traits are generally classified into three categories: those that are always seen (100% of the time), those that are usually seen (over 80% of the time), and those that are frequently seen (between 20% and 80% of the time) (NASF, 1996).

Of the first category, features include:

--severe to profound mental retardation
--profound speech impairment
--movement and balance disorder

**Figure 1**—Patients with Angelman syndrome, displaying classic features

Dr. Angelman’s case 2
Angelman (1965)

Patient Age 50
Reish and King (1995)

Traits that are usually seen include:

--small head size by age three years
--a happy behavior with frequent laughter
--seizures accompanied by an abnormal EEG
--hyperactive behaviors
Finally, in the third category we find:

- flattened back of the head
- protruding tongue and wide mouth
- excessive mouthing and drooling
- strabismus (crossed-eye)
- hypopigmentation of the skin, eyes and hair
- wide-based gait with flat feet
- small hands and feet
- short stature
- heat intolerance
- feeding problems in infancy

Expression of these features aids in clinical diagnosis of the disorder, yet all features need not be present for the diagnosis to be made (NASF, 1996).

Three other features commonly seen in individuals with Angelman syndrome to be addressed in this paper include hypopigmentation of the eye, hypersensitivity and a sleep disorder characterized by a decreased need for sleep.
Ocular hypopigmentation, a congenital, generalized type of hypopigmentation that can be complete or partial, is a commonly seen trait in patients with Angelman syndrome. At this point, little is known conclusively about the connection between hypopigmentation and visual ability. However, hypopigmentation of the eyes is known to effect particular photoreceptors of the retina. "Alteration of the optic system in Prader-Willi syndrome (a similar genetic disorder) is the result of an insufficient amount of melanin present during optic tract development" (King et al., 1993). Results of a test on Angelman syndrome patients to determine the effects of hypopigmentation in Angelman syndrome showed that the fovea light reflex was poor, suggesting abnormal fovea development (King et al., 1993). This is particularly interesting as this information may contribute to behavioral problems in Angelman syndrome. If hypopigmentation in Angelman syndrome does in fact result in reduced or confused vision, it may result further in an inability to focus attention or concentrate on particular stimuli. Should visual and color perception be effected, I believe these behavioral problems may be lessened through use of color perception and preference.

Many Angelman syndrome patients tend to be tactually and visually hypersensitive as well. This may contribute in part to the problem of a short attention span in that these children may be unable to discriminate between different stimuli, and thus unable to concentrate on one or two significant stimuli. The child’s attention is continually averted to new stimuli. In this way, the problems with hyperactivity and attention span are similar to the disorder known as Attention Deficit Disorder. Sensory integration techniques have been effectively used in helping Angelman syndrome patients who suffer from tactile defensiveness, anywhere from waterbeds and bean-bags to deep pressure from tight clothing and body socks. It remains unknown whether hypersensitivity is related to hyperactivity and attention span.

Another physical aspect of Angelman syndrome that may be related to hyperactivity is a sleep disorder characterized by an apparently reduced need for sleep. Dr. Joseph Wagstaff of Harvard Medical Center has been active in researching this aspect of Angelman syndrome. His research has uncovered a link between this sleep disorder and a disturbance of the timing and
quantity of melatonin secretion from the pineal gland. Melatonin supplements have been used therapeutically to help regulate natural melatonin secretion in elderly people. By using .3 mg dose supplements of melatonin, the patients with Angelman syndrome studied by Dr. Wagstaff experienced more normal sleep patterns.

The secretion of melatonin originates in the pineal gland located in the occipital lobe, just anterior to the visual cortex. Melatonin secretion in blind persons is effected by light induction in a similar manner as in people with normal vision. Thus, there may be a connection between visual perception and hormonal balance.

**Diagnosis of Angelman Syndrome**

Clinical diagnosis can be confirmed by genetic testing. Angelman syndrome is caused by a microdeletion on chromosome 15. "Every person has two number 15 chromosomes, one inherited from each parent. In most children with Angelman syndrome, an abnormality can be demonstrated by either directly examining the number 15 chromosomes under a microscope (performing a chromosome study or karyotype) or by using molecular diagnostic methods to examine the chromosomes’ DNA" (Knoll et al., 1991). "However, in 10-20% of Angelman syndrome children, no abnormality has yet been found...Thus, a normal or negative chromosome or molecular study does not necessarily mean that a child does not have Angelman syndrome" (Magenis et al., 1990).
There are generally two laboratory approaches to diagnosing Angelman syndrome (American Society of Human Genetics, 1996). Which approach is used depends on three primary factors: the local availability of testing, previously accomplished studies on the patient, and the level of diagnostic expertise of the referring physician (American Society of Human Genetics, 1996).

Approach 1:
A. Conduct analysis of parent-of-origin status by using southern hybridization with the methylation-sensitive SNRPN or PW71B probes.
   1. If biparental inheritance is identified, most cases of Angelman syndrome are ruled out....
   2. If only paternal alleles are present, the diagnosis of Angelman syndrome is confirmed. FISH and PCR tests can be used to determine whether deletion, UPD or an imprinting mutation is the cause, for genetic-counseling purposes. If the Angelman syndrome is due to deletion, do FISH on the patient's mother to rule out a balanced insertion or an inherited deletion not expressed in the mother.
B. Chromosome analysis should be performed to seek possible chromosome translocation or rearrangement.

Approach 2:
A. Perform chromosome analysis and FISH using SNRPN or other probe in the common deletion region, along with a centromeric probe.
B. Perform methylation analysis, which will detect both UPD and imprinting mutations.
   1. If methylation analysis is normal, most causes of Angelman syndrome are ruled out.
   2. If methylation analysis is abnormal, Angelman syndrome is confirmed only if the paternal band is present.
C. If methylation analysis is abnormal and there is no deletion by FISH, do UPD studies by PCR using micro satellite markers from 15q11-13.
   1. If paternal UPD is present, the diagnosis of Angelman syndrome is confirmed.
   2. If biparental inheritance is identified, an imprinting mutation is suggested. Referral to a research lab should be considered for further molecular investigation.
If the above studies are normal, consultations with a clinician experienced with the disorder is indicated, or a consideration for other conditions with phenotypes similar to Angelman syndrome is suggested (American Society of Human Genetics, 1996).

**Genetic Causes of Angelman Syndrome**

There are four classes of deletions that can occur in Angelman syndrome, as outlined by Dr. Charles Williams at the Canadian Angelman Syndrome Society conference on August 9, 1996. The most common is a deletion of the smallest region of overlap (SRO, seen in 73% of Angelman syndrome cases). It also includes a deletion of the crucial maternal derived Angelman syndrome gene and chromosome translocation. Another type of deletion is what is known as a uniparental disomy (UPD), seen in only about 2% of Angelman syndrome cases. Essentially, the maternal derived chromosome 15 is missing and replaced by a second paternal derived chromosome 15. In fewer that 2% of Angelman syndrome cases, a child has an imprinting (ICR) mutation in which there is an abnormal DNA methylation of 15q11-13. There is also a class, occurring in approximately 23% of cases, in which the mechanism of the deletion is unknown and undetectable by the genetic testing techniques currently used. A fascinating aspect of Angelman syndrome is the fact that when a similar regional deletion occurs on the paternal derived chromosome 15 rather than on the maternal derived chromosome 15, the result is a disorder known as Prader-Willi syndrome. While the disorders are so genetically similar that they were once thought to be identical, and in fact can be diagnosed identically, they are
clinically very different disorders. Prader-Willi syndrome expresses itself in a short, obese stature and compulsive over-eating, sometimes to the extent of eating oneself to death. "For example, a 13 year-old girl...carried 196 pounds on her 4-foot, 4-inch frame!" (Thompson, 1991). This mechanism of inheritance does not appear to follow Gregor Mendel's original genetic principle that identical genes do identical things. "These are the only two syndromes reported in humans with this kind of overlap at this level. The clinical conditions are so distinctive, but the chromosome defects are so similar" (Knoll et. al., 1991).

*Figure 6*—Photo of child with Prader-Willi syndrome

Lai, Erickson and Cassidy (1993)

*Experiences with Individuals with Angelman syndrome*

The subjects used in this project were five individuals with Angelman syndrome; three females and two males. Two females had blond hair and light colored hair, and expressed typical facial and behavioral features of Angelman syndrome. The other girl also expressed typical facial and behavioral features, but had dark hair and light eyes. One of these girls was eight years of age while the other two girls were thirteen years of age. Of the two males, both were blond with light eyes and expressed typical characteristics of Angelman syndrome. One was 24 years of age, and the other was seven years of age.
The project began with observation of normal play activities of these individuals in order to determine color preference. The subjects were each studied individually in their own homes so that interaction could be as normal as possible and they would each feel comfortable. From these observations, it was discovered that all of the subjects’ play preferences were random in that preferences seemed to vary from activity to activity. Specific color differences in toys did not seem to make a difference in the preference of the toy, as long as it did possess some color. Black and white objects were generally not chosen. All of the subjects enjoyed toys that make music, noise or vibration. These toys were usually held close to the ear or near the mouth. Vibrating toys were often used for oral stimulation. In fact, the subjects seemed to put most toys in their mouths. Repetitive play activities appeared to be very stimulating. All of the subjects enjoyed rattling paper.

Because observation seemed to provide little in the way of information on color preference, it was decided to manipulate the environments of the subjects to allow for choices to be made between specific colored items. Two methods were chosen for this experiment. One involved using colored blocks to simply give specific color choices in a play environment as normal as possible. Once again, the subjects were each observed in their own homes. The subjects were assisted in their play in order to manipulate the choices given and record the results. The experimenter arranged a group of blocks consisting of different colored blocks. The colors of blocks involved were yellow, orange, blue, red, and green. A pile of blocks containing blocks of each color was laid out randomly in front of the individual. Most children played on the floor, but one used a table. The subjects were then allowed to play freely with the blocks, choosing one or more blocks of his or her preference. Preference was measured by counting the number of times each color was picked up and manipulated.

It was noticed during the course of this experiment, however, that the subjects responded to the blocks as any other toy, by randomly picking up blocks, sometimes several blocks at once, often not looking at the blocks at all. Once again, as with many other toys, the blocks would go
into their mouths or get thrown on the floor or simply manipulated with their hands. As with the observation, the subjects did not seem to express any preferences for color.

At this point, the second method was chosen. For this experiment, several cards of different colors were presented before each subject. There were two sets of color cards with eight cards in each group, one of eight different colors -- yellow, red, blue, green, orange, purple, black, and white. Each group contained cards of different sizes. One size was approximately seven inches long and five inches wide, while the smaller size was approximately five inches long and three inches wide.

As before, each child was observed in his or her own home. However, for this experiment, each subject was observed at a table or in a quiet environment as free from distractions as possible, rather than simulating a play environment. The cards were presented as a group of eight different colors. The observer held them in her hand in a fan shape, similar to holding a deck of playing cards. The person was then prompted to pick one of the color cards. Preference was determined by which color or colors were chosen more often from twenty-five choices by the subject. The number of choices for each color was counted, and a percentage of choices for each color out of twenty-five was calculated. These results can be found in table 1. This procedure produced somewhat different results in that the subjects did seem to pick certain colored cards more often than others. Each child did seem to prefer or choose a different color, however, thus, the color yellow did not play a significant role in color preference or perception.

**Table 1—results from color preference test**

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Total Trials: 25
The results from the color preference test enabled the study of whether or not color influences attention span. The test to determine whether or not the color of an object plays a role in the length of time an individual with Angleman syndrome pays attention to a task was administered with only two of the original five subjects because of the time involved and the parents of the other three were no longer willing to participate. The test was run with the two remaining subjects. The question at hand was whether or not a particular color of an object or stimulus could have an influence on an individual’s attention span. Because the color cards produced the best results when testing for color preference, the cards were again used to test for attention span. The subjects were given the cards one at a time in random order, and the time spent looking at, playing with touching or mouthing each card was recorded using a watch with a timer. Subjects were timed from the moment the individual picked the card until when the card was put down or handed back to the observer. The objective of this portion of the project was to discover whether or not the color of an object would have an effect on attention span towards a task by eliciting a length of attention greater than one minute. It was concluded from the results, however, that color preference appears to have little if any effect on the length of attention span. The preferred color for each of the two girls did not appear to lengthen attention span towards the cards, nor did any color presented. Rather, differences in attention span seemed to be due to some other stimulation; for example, a noise or visual stimulus which would cause the child to focus her attention span on something else. In addition, attention span actually was less under the experimental conditions than under natural play conditions, probably due to the creation of an unnatural environment, which can be distracting for a child with Angelman syndrome. In other words, when placed at a table and asked to choose a card or to play with blocks, these children were more easily distracted than if they were allowed to participate in an activity of their choice in a location of their choice. This seems to be normal. One’s attention span is likely to be greater when attending to a task that is enjoyable and freely chosen.
Discussion

As stated at the beginning of this paper, the reason for my interest in Angelman syndrome came from experiences working as a care-provider for children with the disorder. Due to a curiosity about Angelman syndrome and in an attempt to learn more about the children that I work with, I began writing to parents of children with this disorder via the internet. As I communicated with parents, one mother mentioned a break-through her child had. She said, "About a year ago, we bought the balls from McDonald’s that you jump in. The night we got them, we walked into the room and saw that she was sorting out all the yellow balls!!! This was the first cognitive/academic thing we’d seen...." Other parents responded by saying that some of their children seemed to have favorite colors, most of them yellow. I began to think that if this child was attending to the task of sorting her favorite color, perhaps that color could help her focus her attention on other tasks as well. With this in mind, I decided to test for color preference amongst children with Angelman syndrome. Due to the fact that several other parents responded also indicating that their children seemed to prefer yellow to other colors for whatever reason, I became curious as to whether there was a common color preference amongst these children. Another example comes from Jeanne Seltzer, whose son has Angelman syndrome. As she was talking to one of her son’s doctors, he asked if he had a preferred color. When she responded “yes,” he asked, “Let me guess, it’s yellow, right?” This indicates that he has seen other children with Angelman syndrome who displayed a preference for the color yellow. It is interesting to note that a few parents also mentioned other colors, but one thing remained the same. Each of these children responded strongly to one specific color. If these tests resulted in a positive color preference, then I could also test to see if that color played any role in the child's attention span. This project is devoted to the behavioral aspects of Angelman syndrome that are addressed in this paper. The biological components of the disorder that may relate to behavior are also fascinating and deserve further investigation to help determine if there are other possible methods of alleviating behavioral and additional problems associated with Angelman syndrome.
Angelman syndrome is still a rare disease, and while many genetic and health concerns have been addressed, there has been little research or literature concerning the behavioral problems associated with Angelman syndrome. Hyperactivity and a short attention span can be behavioral nightmares for any parent, and it is no different for parents of children with Angelman syndrome. The major exception is the incredibly short length of attention span for these children and their apparently decreased need for sleep as described above. Most parents of children with this disease would give almost anything to help their children and their families alleviate this problem. Training is often very difficult, if not impossible, and communication can also be difficult and disheartening at times. I and the people that I have worked with understand these feelings. That is the importance of beginning this work.

If this project could be done again, I would suggest modifying the methods. The methods used were not appropriate for measuring attention span in people with Angelman syndrome. The children seemed more prone to distraction during the time that data was being collected. Knowing one of them quite well, and using the knowledge of the others’ parents, it was obvious to me that these children have a much longer attention span when involved in a task of their own choosing. Short attention span seems to be more of a problem when asked to participate in an activity in which he or she is not interested. In this way, these children are not different from anyone else. Of course, one’s attention span is greater when involved in something interesting or enjoyable!

The most important thing gained from the experience of conducting this research and obtaining the knowledge from it is that these children with such a rare and unique disorder, while they seem so different, they really are not. They possess many of the same qualities as normal people, and communicate the same needs and desires. It is up to those of us that are normal to listen to what they can tell us.
Photos of Some Special Children

Kyle

Richard

Lacey

Andy

Mikey
REFERENCES


