

DEVELOPMENTAL GERSTMANN'S SYNDROME

A CASE REPORT & LITERATURE REVIEW

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Abstract

Developmental Gerstmann's syndrome is a neurological disorder in children characterized by a tetrad of symptoms including dysgraphia, dyscalculia, left-right confusion, and finger agnosia. The incidence and exact cause is unknown. Controversy surrounds the existence, diagnosis and treatment of this condition. The case presented here represents a patient with developmental Gerstmann's syndrome that was subsequently diagnosed with visual efficiency difficulties and visual perceptual deficiencies. A literature review is included.

Key Words

accommodative insufficiency, directionality, dyscalculia, dysgraphia, finger agnosia, Gerstmann's syndrome, laterality, perceptual deficiency

INTRODUCTION

Gerstmann's syndrome (GS) is a neurological disorder that was first described in the 1930's,¹ and has been subsequently characterized by four signs: dysgraphia, dyscalculia, left-right confusion, and finger agnosia.²⁻¹² Controversy exists as to whether GS represents a true syndrome or a collection of symptoms that often occur together. There is a relative absence of reports of this condition in the literature. It is reported to occur in both adults and children.²⁻⁴ In adults, the condition is typically acquired (AGS) and may occur after a stroke, in association with damage to the cortex,^{2,6} or in Alzheimer's disease.⁹ The condition is termed developmental Gerstmann's syndrome (DGS) when these symptoms coexist in a child.²⁻¹²

While definite causes have been identified in adults, the same cannot be said concerning children. Language deficits that present in AGS are not found in children.¹³⁻¹⁵ Conversely, reading impairment that occurs frequently in DGS is not observed in AGS.^{13,14,16} In fact, controversy surrounds the very diagnosis of DGS. There is little consensus as to how to make a proper diagnosis as well as the specific tests required. The amount of specific deficits needed to make a diagnosis is also controversial.²⁻⁴ Another compounding factor is that several of the tests do not have norms.³ It is unclear whether the tetrad of findings associated with DGS (Table 1) requires that all four associated symptoms be present for a diagnosis to occur, or if partial expression of the findings is possible.

Signs of DGS

Finger agnosia (aka finger aphasia), one of the four conditions found in DGS, was characterized by Gerstmann. His description was "a loss of the ability to recognize,

identify, differentiate, name, select, indicate and orient as to the individual fingers of either hand, the patient's own, as well as those of another person."^{17,18} The developmental age when finger naming occurs remains controversial.²

Dysgraphia, a writing disability, is characterized by errors in spelling and penmanship. This sign is also tricky to decipher.³ The most common spelling errors found are related to the letter sequence, as well as omission, substitution and transposition errors.^{13,15,19} Penmanship issues are described as poor letter formation, letter orientation and letter part orientation. Writing is poorly aligned and demonstrates a poor use of space.³

Dysgraphia in DGS is attributed to constructional apraxia, a fifth proposed characteristic of the syndrome. Constructional apraxia is an inability to reproduce simple drawings of shapes, and impairment in tasks that involve constructing patterns using colored blocks. Poor performance in handwriting and constructional tasks is interpreted as representing difficulties in understanding spatial concepts.²⁰

Acuculia or dyscalculia is a lack of understanding of the rules for calculation or arithmetic.^{3,4} Deficiencies have been noted with DGS in both verbal and written math calculations.¹⁵ According to one study, children with DGS understand the concepts that form the basis for the math calculations but have poor ability to write and sequence numbers.²⁰ A contrasting opinion puts forth that the math errors are "due to a Piagetian developmental delay as the child transitions from the preoperational to the concrete operational stage of development."⁵

The fourth characteristic of DGS is the inability to distinguish right from left on one's self as well as on another person. This is known as left-right confusion or

Table 1. Findings of Developmental Gerstmann's Syndrome

Diagnostic Findings	Finger agnosia Left-right confusion Dysgraphia Dyscalculia Constructional apraxia*
Associated Findings	Average or better than average intelligence Elevated verbal intelligence Negative family history for learning disabilities Appropriate language development Behavioral problems Gait abnormalities Increased muscle tone Hyperactive tendon reflexes Extensor plantar responses Tremors

*Hypothesized diagnostic finding

a laterality deficit.³ Studies have offered varied descriptions in this area ranging from slowness or hesitancy in responding,²¹ to being unable to follow directional navigation instructions during daily activities.¹⁶

Proposed Neurology

The exact cause of DGS is unknown but there are several models and hypotheses. Specific brain areas that have been implicated are the parietal lobe,¹³ and a sub-angular lesion. The latter lesion is a focal ischemic lesion, situated sub-cortically in the inferior part of the left angular gyrus and reaching to the superior posterior region.^{7,8} While it is theorized that parietal lobe damage to either side is implicated,²⁰ the left-hemisphere angular gyrus²² and the right supra-marginal gyrus²¹ may be responsible. Although here are no specific neurological abnormalities documented in the literature,³ a recent study did find abnormal EEG recording on four out of ten patients. Nonspecific findings were documented, such as hyper-intense lesions in the white matter and empty sella syndrome. This syndrome is a radiological finding where spinal fluid is found within the space created for the pituitary²³ It is yet unknown if these abnormalities are related to DGS.²

Abnormal developmental processes causing neuro-cognitive skill abnormalities have been considered as potential causation of DGS. Kinsbourne¹⁷ suggested that developmental delays were the cause of DGS and that an underdeveloped "neural facility" or perinatal trauma caused a subtle manifestation of cerebral palsy. Researchers have not located an area in the brain responsible for the symptoms seen in DGS. The search for a specific area has proven difficult, as the multiple skills

deficits shown in DGS are controlled by, and therefore affected by, multiple brain loci.³

The incidence of DGS is likewise unknown. A literature search on PubMed reveals a paucity of case reports and no epidemiologic studies. It is unknown if there is a gender or race predilection. Familial occurrence has also not been documented with DGS.¹⁵ Many consider DGS to be rare, others feel that it may be more common than reported.⁴ Lack of awareness of the condition, unreliable or unperformed testing procedures, the overshadowing by other conditions, as well as behaviors being attributed to other conditions, are all cited as potential reasons for a low rate of diagnoses.⁴

The diagnosis of DGS itself is compromised. The tests used to diagnose the condition are not standardized, nor are norms by age and gender available. For example, in making a diagnosis of finger agnosia, four types of tasks are assessed.

- 1) Identification tasks can be evaluated by showing the examiner which finger was touched while blindfolded and then naming the finger.^{22,24}
- 2) Differentiation tasks that require the child to identify whether the examiner touched one or two of their fingers³
- 3) "In-between tasks" require the child to identify the number of fingers between two that were touched.
- 4) Finger block tasks use blocks that are shaped so that the fingers form a specific pattern. The child's hand is closed around a block that elevates some of the fingers. The block is removed, and the eyes that were previously closed are opened. The child is asked to identify which block out of a group of blocks they were holding.³

Of the studies published, the test protocol was not standardized. Some clinicians reported one type of task used in the diagnostic battery, while others use a combination.³

While those diagnosed with DGS exhibit the diagnostic criteria, there are associated features that have been documented in the literature. Average or better than average intelligence, as well as elevated verbal intelligence, have been noted in some reports.³ As much as a 20-point split has been documented when comparing verbal to performance scores on the Wechsler Intelligence Scale for Children (WISC).¹³ This is the primary measure of intelligence used in the United States. Also, most studies report a negative family history for learning disabilities or inappropriate language development.³ Behavioral problems have been found in greater numbers in those diagnosed with DGS. These include attention-deficit-hyperactivity disorders and emotional disturbances such as temper tantrums, aggressiveness, spitting and biting.² Several "soft" neurologic signs have also been reported in children with DGS. Gait abnormalities include unusual and uncontrollable problems with walking due to conditions such as vestibular and central nervous system disorders.²⁵ They may also show: increased muscle tone, hyperactive deep tendon reflexes (abnormally brisk muscle contraction occurring with a sudden sharp tap to the muscle's tendon of insertion),²⁶ extensor plantar responses (abnormal reflex of dorso-flexion of the great toe and abduction of the other toes when the plantar surface of the foot is stimulated),²⁷ and tremors (rhythmic, involuntary, oscillatory movement of body parts).²⁸

Similar Conditions

There are several well known syndromes that have been documented as associated with DGS. In one study 65% of individuals with Fragile X syndrome demonstrated three or more of the diagnostic characteristics of DGS.²² Williams syndrome, a rare genetic condition (~ 1/7,500 births), has both similar and contrasting symptoms of DGS. Affected individuals with William's syndrome exhibit visuo-spatial construction problems with elevated verbal intelligence.²⁴ They have symptoms of inattention and below average performance intelligence, but display strong auditory short term memory.²⁹

Patients with nonverbal learning disabilities (visuo-spatial learning deficits, math calculation deficits and deficits with tactile perception) and Asperger's syndrome also have both similar and dissimilar symptoms to DGS. Unlike DGS however, these two conditions also present with issues relating to nonverbal problem solving, pragmatic speech, and poor spatial judgment.² Those with Asperger's syndrome characteristically show evidence of poor visuo-spatial skills, normal intellectual functioning, elevated verbal intelligence, math deficits, and right hemisphere deficits. In contrast to DGS, they display poor social interactions, circumscribed interests and poor nonverbal communication.³⁰

Since the number of reported cases of DGS has been minimal, determination as to the natural history is not well known. Both full and partial resolution has been documented in the literature.⁴ Even cases of spontaneous resolution have been reported, but some evidence of a learning disability remained.¹³ Given that adult and teenage cases have been detailed, it is evident that some cases persist and are resistant to remediation.¹⁶

Medical and educational treatment is typically symptomatic and supportive. Reading tutoring for dyslexia or graphomotor training for dysgraphia has shown to be mostly unsuccessful. Treatment traditionally includes occupation and speech therapy.⁴ Compensatory learning techniques such as calculators and computer word processors, bypassing the deficit when possible, may prove to be more effective in the long run. Most importantly, the patient, parents and teachers must be educated on the condition and the possible secondary limitations. This is believed to help alleviate and prevent exacerbation of already existing emotional difficulties.⁴

CASE REPORT

History

A nine-year-old male presented with complaints of difficulty in school. Currently in fourth grade, he has been receiving speech therapy and attending extended resource special education classes since kindergarten. The patient's mother indicated that he has difficulty completing assignments particularly in reading, writing, arithmetic and spelling.

The patient was delayed in motor and language milestones. The first words were spoken at 2 years of age and two to three word sentences at age 4. The patient has

Table 2. Optometric examination data

Unaided Visual Acuity- Distance Near	OD 20/20 OD 20/20-	OS 20/20 OS 20/20-	OU 20/20 OU 20/20
Cover Test-Distance Near	Orthophoria 8 exophoria		
Stereo	250 sec arc global, 70 sec of arc local (Randot)		
Distance Retinoscopy	OD -0.25-0.25 X 090 OS Plano	20/20- 20/20-	
Manifest Refraction	OD PL-0.25 X 090	20/20	OS Plano 20/20
Phoria-Distance Near	Orthophoria 2 esophoria		
NRA/PRA	+2.25/-1.00		
Near Vergences	BO 12/24/12	BI	16/20/12
MEM	OD +1.00 OS +1.00		
Accommodative Amplitude	OD 9.00D OS 9.00D		
Accommodative Facility	OD 10cpm (+) harder OD, OS; (-) harder OU	OS 9cpm	OU 7cpm
NPC	1)13/17 2)17/19 3)17/20		
NPC with probe lens (+0.50D)	1)5/7 2)5/10 3)6/8		

BO=Base out

BI= Base in

cpm= cycles per minute

MEM= monocular estimate method

NRA= negative relative accommodation

PRA= positive relative accommodation

not learned to tie his shoe, nor ride a bicycle or tricycle. Toilet training was also delayed, occurring at 3½ years of age. It was reported that the patient has difficulty relating to others his own age as well as being immature and clumsy.

While the pregnancy was uncomplicated, labor was induced due to toxemia. No postnatal complications were reported and the child did not suffer from unusual childhood illnesses. His medical history was positive for multiple ear infections. The patient did not use medications. An uncle was reported to have a seizure disorder and hearing problems, but the patient's direct family medical history was free of significant medical or emotional problems.

Two years prior, the patient had been diagnosed with Attention Deficit Hyperactivity Disorder (ADHD), Gerstmann's syndrome and upper body weakness by a neuro-psychologist. That evaluation revealed low average cognitive processing abilities and below average achievement skills. His gross visual-motor integration ranged from severely impaired to average. He exhibited low to mildly impaired receptive vocabulary abilities. Impairments in visual memory, executive functioning and symbolic transfer (transformation of non-verbal signs into verbal representations) were also noted. The Wechsler Abbreviated Scale of Intelligence (WASI) indicated a full scale IQ of 87. Treatments

outlined included medical therapy for ADHD, as well as academic strategies and modifications to cope with the deficits.

Optometric Findings

A COVD-Quality of Life-Short Form (COVD-QOL)³¹⁻³⁵ was completed producing a score of 41. A score of greater than 20 is considered indicative of a binocular vision dysfunction. The data (Table 2) was obtained at the initial examination. Looking closely at the accommodative data (low PRA, high NRA, low accommodative amplitudes, lag on MEM) a pattern of accommodative insufficiency is revealed. While the reduced Near Point of Convergence (NPC) can indicate convergence insufficiency, retesting with a low plus lens negated this finding. The internal and external structures of the eye were within normal limits. The patient was diagnosed with an accommodative insufficiency and pseudo-convergence insufficiency. The patient was issued a near prescription of +0.50D for use in school and at home. Due to the high score of the COVD-QOL questionnaire, as well as the patient's history of school difficulties, a perceptual examination was performed following several weeks of wearing the near prescription.

A perceptual examination (Table 3) was performed at a later date which indicated a deficiency in laterality and directionality. Dysgraphia was not diagnosed due to at least average performance on two

Table 3. Perceptual examination data

TEST	CLASSIFICATION	PERCENTILE/ AGE EQUIVALENT	Rank
Piaget Right/Left Awareness test	Laterality and Directionality	Age 6 years	Very Weak
Standing Balance	Assesses body schema and motor planning	< Age 4.0-4.5	Very Weak
Six Figure Split Form Board	Assesses simultaneous processing and eye-hand coordination	80 %	Strong
Gardner Reversal Frequency Test --Execution	Requires child to write numbers & letters as called out in random order	1%	Very Weak
Gardner Reversal Frequency Test --Recognition	Requires child to recognize letters & numbers written backwards/reversed	<1%	Very Weak
Gardner Reversal Frequency Test --Matching	Evaluates visual attention and discrimination	<17%	Weak
Developmental Eye Movement Test (DEM): Horizontal	Developmental Eye Movement test: Saccadic eye tracking used for reading	57%	Average
(DEM) Vertical		62%	Average
(DEM) Ratio		46%	Average
(DEM) Errors		76%	Strong
The Beery-Buktenica Developmental Test of Visual-Motor Integration (VMI)	Visual Motor Integration -simultaneous processing -eye-hand coordination	42%	Average
VMI-Motor Coordination		39%	Average
Wold Sentence Copy Test	Requires child to copy a paragraph -Eye hand coordination -Fine Motor Skills -Eye tracking	> 8th grade	Very Strong
Dyslexia Determination Test (DDT) Eidetic coding	Dyslexia Screening Instrument	Above Normal	Very Strong
Dyslexia Determination Test (DDT) Phonetic coding		Above Normal	Very Strong

paper and pencil tests for visual motor integration (Wold Sentence Copy^a and Beery-Butenica Visual Motor Integration^b). Dylexia testing performed with the Dyslexia Determination Test (DDT)^c showed above normal eidetic and phonetic coding. The patient's highest grade level of sight words recognized was 7-8th. This is three to four grades higher than the patient's current grade level. The Developmental Eye Movement Test (DEM),^{36,d} revealed no deficit in ocular motility or automaticity.

The patient was entered into a vision therapy program to address both visual skills (accommodative insufficiency) and perceptual deficiencies. It was estimated that 15-20 visits would be needed to remediate the accommodative aspects while 30-40 visits would be needed for laterality and directionality difficulties. Unfortunately, the patient's mother was unable to continue his scheduled therapy after completing only five visits over a period of 15 weeks. The patient was scheduled to be re-evaluated during his yearly scheduled vision examination.

DISCUSSION

This case represents a child diagnosed with a neurological disorder that closely resembles a visual efficiency (accommodative dysfunction) and perceptual dysfunction. Looking closer at our perceptual testing results in relation to the diagnostic criteria for DGS, there are both similarities and dissimilarities. Based on tests such as Piaget Right/Left Awareness Test^a and Gardner Reversal Frequency Test^a the patient was diagnosed with deficiencies in laterality and directionality and poor coding of words with left-right confusion. These signs are basic tenets of DGS. In contrast, testing in the area of visual motor integration, a component of dysgraphia, was not deficient. Our patient did not show any trouble in any of the copying activities. The Wold Sentence Copy^a showed proper letter formation, adequate spacing and no letter reversals. Visual motor integration and motor coordination testing both showed results in the average range, negating another diagnostic criterion, constructional apraxia. Finger agnosia and dyscalculia are not areas traditionally tested via optometric perceptual examination so no comparison can be made in these areas.

Why did our testing show a difference in two out of three areas diagnostic for DGS? One explanation is that the testing performed previously under the supervision of the neuro-psychologist occurred several years prior to the perceptual testing.

Cases of partial resolution have been documented in the literature,^{4,13} perhaps this case fits into that category.

Another possibility is that perhaps the patient simply had perceptual deficiencies and visual efficiency issues at the time of the initial diagnosis. While it is impossible to travel back in time to suggest a referral to an optometrist who specializes in learning-related vision problems; the lack of vision care in this child and many others in documented cases is disappointing.^{2,4,6,7,9,11,12} Many of the diagnostic signs are, in fact, related either directly or

indirectly to the visual system. It is possible that children diagnosed with DGS are actually children with significant visual skills and visual information processing disorders. Certainly, the care of a behavioral optometrist should be considered in any diagnosis of DGS.

CONCLUSION

This case report details a patient diagnosed with developmental Gerstmann's syndrome. The main difference in this case versus others documented in the literature is that this report is the first to demonstrate a visual efficiency problem in a patient diagnosed with DGS. Even though many of the symptoms of DGS are vision related, the need for an examination of the visual system has not been documented in the literature and therefore may not be recommended or encouraged. The question remains as to the prevalence of visual efficiency problems in those with DGS and whether these patients may simply have undiagnosed and untreated visual problems. Regardless of the cause of the visual problem, the behavioral vision care profession can potentially make a huge contribution to the emotional, social and academic well being of these children. Behavioral optometry must be at the forefront of the treatment of these children.

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