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Developmental and Persistent Developmental Stuttering: An Overview for Primary Care Physicians

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Stuttering is a speech disorder characterized by a disruption in the fluency, timing, and rhythm of normal speech. It affects approximately 5% of children at some point in their lives. Although dysfluency often resolves before adulthood, it may cause periods of extreme anxiety for patients, especially those who continue to stutter in adolescence and adulthood. Although these patients are unlikely to stop stuttering, treatment options are available to reduce anxiety and therefore the severity of symptoms. In the present review article, the authors discuss the pathophysiology, diagnosis, and management of developmental stuttering in children and adults.

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Stuttering occurs in people of all ages, ethnicities, and cultures, but it is most commonly associated with young children as they develop and learn language and speech.[1] Approximately 5% of all children will experience some form of speech dysfluency.[2] However, studies have shown that 50% to 80% of those who stutter will recover spontaneously by puberty without the intervention of professional treatment.[3]

As defined by the *Diagnostic and Statistical Manual of Mental Disorders*,[4] stuttering is "a disturbance in the normal fluency and time patterning of speech that is inappropriate for the individual's age." Stuttering manifests itself as repetitions of sounds, syllables, or words or as a speech block with prolonged pauses between sounds and words. Secondary behaviors such as eye blinking, jaw jerking, and head movements are learned approaches to minimize the severity of stuttering and can lead to increased fear of speaking and embarrassment.[5,6] Adults who stutter often develop linguistic escapes and behaviors such as word substitutions, interjections, and sentence revisions.[5,6] Because of these behaviors, stuttering has been shown to interfere with academic and professional achievement, as well as social communication.[6]

Stuttering has been classified as developmental, neurogenic, and psychogenic (*Figure 1*).[6] Developmental stuttering (DS) is the most common form and encompasses all cases with a gradual onset in children, generally between the ages of 3 and 8 years.[1,5,6] The term *developmental* is used because this form of stuttering occurs during the period of extensive speech and language development.[5] Speaking in front of a group and talking on the telephone tend to worsen DS, while singing, reading aloud, and speaking alone relieve dysfluency.[4] According to Yairi and Ambrose,[7] approximately 75% of preschoolers with DS undergo spontaneous remission within 4 years.

Persistent DS is a form of DS that has not resolved, either spontaneously or from speech therapy.[5] The Stuttering Foundation of America[2] reports that 1% of the global population stutters. In the United States, that percentage is equivalent to more than 3 million people.

Type of Stuttering	Definition
Developmental	Stuttering with a gradual onset during childhood; presents as a dysfluency in the timing, patterning, and rhythm of speech
Neurogenic	Typically the result of nerve or traumatic brain injury
Psychogenic	Begins suddenly after emotional trauma or stress; also occurs in patients with history of psychiatric illness

Figure 1. *Classifications of stuttering. Developmental stuttering that has not undergone spontaneous or therapy-induced remission is often referred to as persistent developmental stuttering.*

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Neurogenic stuttering is rarer than DS and typically occurs after a brain injury event (eg, traumatic brain injury, stroke, Alzheimer disease).[5,6] Neurogenic stuttering is easily differentiated from DS because patients with neurogenic stuttering usually lack secondary behaviors.[5,6]

Psychogenic stuttering is another rare form of dysfluency. It is characterized by the rapid repetition of the initial sounds of a word.[8] It usually occurs in adults with a history of psychiatric illness or after emotional trauma.[8]

In the present article, we discuss the pathophysiology, diagnosis, and management of DS and persistent DS. We also review behavioral therapy and pharmacologic treatments that have shown promise in the literature.

Pathophysiology

Evidence supports a direct link between genetics and stuttering. Family and twin studies[9,10] have all shown a direct correlation to stuttering and inheritability; in 2000, Felsenfeld et al[9] studied twins and found that 70% of DS was linked to genetics.[9] More recently, a missense mutation in the *N*-acetylglucosamine-1-phosphate transferase gene (GNPTAB), found on chromosome arm 12q, was linked among families in Pakistan.[10] The researchers[10] hypothesized that this variant caused lysosomal malfunction in which the efficiency of lysosomal targeting of catalytic enzymes is reduced. However, further research is needed to draw a firm conclusion.[11]

The sex of the patient plays a key role in the development and even the persistence of stuttering. The male-to-female ratio of DS in children is 2 to 1; in adults with persistent DS, the ratio can be as high as 5 to 1.[7] Review articles[5,6] have noted that stuttering is more likely to resolve in females than in males, and males are more likely to develop persistent DS. Of men with persistent DS, 9% of their daughters and 22% of the sons will go onto develop DS.[12] However, when compared to women who have persistent DS, 17% of their daughters and 36% of their sons will develop DS.[12]

Studies have shown that adults with persistent DS have different cognitive processing abilities compared with adults with normal speaking behaviors. Several organic models have shown incomplete lateralization and abnormal cerebral dominance in those who stutter.[5,6] In fluent speakers, the left hemisphere of the brain, which is language-dominant, is more active during the speaking and language processes.[1,13] However, in speakers with DS, the right hemisphere appears to be more active in the language process but the left hemisphere is more active in the production of stuttered speech.[13] In people with fluent speech, early activation occurs in the left frontal brain, which involves language planning; the activation occurs before the central areas become involved in speech.[1,13] However, in individuals with DS, this process appears to be either absent or reversed.[13]

Not only have cognitive processes been linked to DS, but also key structural abnormalities have been noted. The first anatomic variations seen in individuals who stutter were abnormalities in both the Broca convolution and Wernicke area of the brain—areas that support speech and language.[14] The researchers also found abnormalities in gyrification patterns, which support the idea that DS occurs during early development of the brain.[14] Furthermore, those with DS have decreased white matter tract coherence in the Rolandic operculum.[15] This area of the brain lies adjacent to the primary motor cortex of the larynx, pharynx, and tongue and the inferior arcuate fascicle, which links the temporofrontal language system.[15] Because of these regional relationships, Sommer et al[15] hypothesized that the fast sensorimotor integration necessary for fluent speech is disrupted in individuals with DS.

Although structural abnormalities and cognitive processes have been linked to DS, a new hypothesis is that DS is a speech disorder resulting from a central neuromotor dysfunction involving dopamine receptors that disorganize the exact timing needed to generate fluent speech.[5,16] For example, Costa and Kroll[5] report the following in their review article:

> Positron emission tomography studies using 6-fluorodopa as a marker of presynaptic dopaminergic activity showed significantly higher [6-fluorodopa] uptake in patients with moderate to severe DS than in non-stuttering control subjects.

Specifically, uptake was greater in the ventral limbic cortical and subcortical regions, which are strongly associated with the modulation of speech.[5]

The serotonin systems in patients with DS appear to have a more restricted role than in those with fluent speech and appear to be linked with the metabolism of dopamine. Moreover, the serotonin system has been linked to intracortical excitation and increasing the cortical silent period.[5] However, further research in this area is needed to support these conclusions.

Diagnosis

Developmental stuttering will typically occur before the age of 12 years.[17] However, many preschool-aged children undergo a dysfluency period that makes it hard to distinguish DS from other types of dysfluency.[5] As a result, the Stuttering Foundation of America[2] created a risk factor checklist that can be used by parents to gauge when to seek treatment for their children from a speech therapist (*Figure 2*).

The initial assessment of a patient suspected of having DS should be based on determining the severity of the dysfluency, family history, and concern about stuttering behaviors (*Figure 3*). The Stuttering Foundation of America[2] currently breaks down dysfluencies into normal dysfluency, mild stuttering, and severe stuttering. A patient with normal dysfluency will present between ages 1½ and 3 years, and the dysfluency is differentiated by brief repetition of sounds and words at the beginning of a sentence.[2] In normal dysfluency, stuttering

Risk Factor	More Likely in Beginning Stuttering	True for My Child
Family history of stuttering	Parent, sibling or other family member who still stutters	☐
Age at onset	After age 3½ years	☐
Time since onset	Stuttering 6-12 months or longer	☐
Gender	Male	☐
Other speech-language concerns	Speech sound errors, trouble being understood, difficulty following directions	☐

Figure 2. *A chart of risk factors of stuttering for parents. These risk factors place children at higher risk for developing stuttering. If a child has shown signs of stuttering and meets any of these risk factors, the parents should consult a speech-language pathologist who specializes in stuttering. Reprinted with permission from the Stuttering Foundation of America.*[2]

occurs once in every 10 sentences.[2] Moreover, children with a normal dysfluency will have little or no frustration or any awareness of their speaking disabilities.[2]

Mild stuttering is associated with children aged 3 to 5 years.[2] Although patients with mild stuttering present similarly to those with normal dysfluencies, several key characteristics can differentiate the 2 conditions. Mild stuttering may be accompanied by secondary behaviors, and the frequency of the dysfluency is more prominent.[2] Also, some anxiety and embarrassment in the child is usually noted with mild stuttering.[2] These patients should be referred to a speech-language pathologist if symptoms persist for more than 6 weeks.

Severe stuttering is typically seen in children aged between 1½ and 7 years.[2] The patient's stuttering will occur in less than 20% of their spoken words but will occur in nearly every sentence.[5] The patients will have numerous secondary behaviors associated with their speaking and will also show numerous characteristics of avoidance, especially fear of speaking.[2] When treatment is started early in this subset of patients, outcomes tend to be better.

Treatment

Untreated stuttering has been known to cause several debilitating physical and psychosocial phenomena. Physically, those who stutter report tense musculature. Socially, 70% of adults who stutter feel they are adversely affected at their job, while 20% have reported declining a promotion at work because of their dysfluency.[18] Thus, stuttering evokes not only fear, anxiety, and embarrassment but also a decline in self-esteem and self-perception. Although there is no cure for stuttering, it is important for physicians to be aware of current treatment options for patients who stutter.

Nonpharmacologic

Speech therapy remains the first-line treatment method of choice by most physicians.[2] Unfortunately, patients who are in the classification of severe stuttering or are older than 18 years will see little or no results with intensive speech therapy.[5,6]

Speech therapy has evolved into focusing on decreasing symptoms of secondary behavior and managing stuttering events instead of the more traditional approach of trying to cease the speaking dysfluency. The focus of speech therapy is to halt progression of the dysfluency while teaching the patient how to effectively manage his or her disorder.[19] Venkatagiri[19] has shown that this method minimizes the impact and occurrence of stuttering but does not completely eliminate it from everyday life.

A second nonpharmacologic management mechanism is the fluency-shaping method, which relies on a delayed auditory device. In this method, the rate of speech by a speaker must slow to prevent heard distortions through the electronic device.[20] This has been known as the choral effect, and its efficacy is directly related to the severity of the dysfluency.[20]

The Lidcombe approach[21] has been effective in preschoolers with stuttering. During the treatment phase of a study, parents use a form of operant conditioning to enhance the child's fluency. Parents provide their child with an environment that encourages the child to speak relatively slowly.[21] The child is then praised for fluent speech but is not discouraged when dysfluency is present.[21] Instead, occasional corrections are used to steer the child back toward fluency.[21]

Although most mild cases of DS regress with age and speech therapy, if a patient presents with DS past the age of 8 years, elimination of stuttering substantially decreases.[7] In patients older than 8 years, 2 treatment methods are typically used. The goal of the first method is to shape the patient's fluency from stuttering to normal pattern speaking.[6] This is accomplished by the patient controlling the rate and rhythm of his or her speech through constant self-monitoring techniques.[6] The goal of the second method, which is referred to as stuttering modification, is to reduce the fear associated with overt stuttering and decrease primary and secondary behaviors.[6] Stuttering modification is accomplished through attempts at reshaping the respiratory, phonatory, and articulatory gestures used to generate speech.[6]

Pharmacologic

Costa and Kroll[5] and Prasse and Kikano[6] conducted reviews of the literature on pharmacologic treatment options to control or reduce stuttering and found that medications were ineffective or had deleterious effects on patients. Currently, there

Checklist Item	Normal Dysfluency	Mild Stuttering	Severe Stuttering
Speech behavior you may hear or see	Occasional (not more than once in every 10 sentences), brief (typical ½ s or shorter) repetitions of sounds, syllables, or short words	Frequent (3% or more of speech), long (½ to 1 s) repetitions of sounds, syllables, or short words; occasional prolongations of sounds	Very frequent (10% or more of speech) and often very long (1 s or longer) repetitions of sounds, syllables, or short words; frequent sound prolongations and blockages
Other behavior you may see or hear	Occasional pauses, hesitations in speech, usage of filler words, or changing of words or thoughts	Repetitions and prolongations begin to be associated with eyelid closing and blinking, looking to the side, and some physical tension in and around the lips	Similar to mild stutterering only more frequent and noticeable; some rise in pitch of voice during stuttering; extra sounds or words used as "starters"
When problems are most noticeable	Tends to come and go when child is tired, excited, talking about complex or new topics, asking or answering questions, or talking to unresponsive listeners	Tends to come and go in similar situations but is more often present than absent	Tends to be present in most speaking situations; far more consistent and nonfluctuating
Child reaction	None apparent	Some show little concern, some will be frustrated and embarrassed	Most are embarrassed and some are also fearful of speaking
Parent reaction	None to a great deal	Most concerned, but concern may be minimal	All have some degree of concern
Referral decision	Refer only if parents are moderately to overly concerned	Refer if continues for 6 to 8 weeks or if parental concern justifies it	Refer as soon as possible

Figure 3. *Physician checklist for children with speaking dysfluencies. Age of onset is between 1½ and 7 years. Reprinted with permission from the Stuttering Foundation of America.*[2]

is no pharmacologic agent that will reduce stuttering to less than half of the prior frequency or to decrease the dysfluency to less than 5% of spoken words.[6] As a result, no pharmacologic treatment can be endorsed by the US Food and Drug Administration; however, several medications have shown promise in decreasing the amount of stuttering through research based on the dopamine and serotonin models.

Maguire et al[22] found that risperidone (2 mg/d) can be used in patients with moderate to severe stuttering with minimal adverse effects. Risperdone works as a dopamine antagonist to effectively decrease the amount of dopamine in several key areas of the brain and theoretically lead to an increase in modulation of normal language.[22] Although, Macguire et al[22] showed that risperdone lowers the amount of dysfluency to less than 5% in their study, this appears to be compromised by the fact that the placebo group also had a decrease in dysfluency to 5%.

Olanzipine, an atypical antipsychotic drug, has also been implicated in decreasing the amount of stuttering in patients diagnosed with DS. In a double-blind placebo-controlled study from 2004, Maguire et al[23] reported that when olanzipine was titrated from 2.5 mg to 5 mg each day, participants with DS had a significant decrease in dysfluency compared with participants in the placebo group.

Also, pagoclone, a nonbenzodiazepine γ-aminobutyric acid modulator, has shown a reduction in percentage of syllables stuttered when compared to placebo.[24] The most common adverse effect was headache, which was reported in 12% of patients who received 0.3 mg of pagoclone twice a day.[24]

In adults, persistent DS is usually associated with either depressive symptoms or social phobia with a general anxiety disorder. Research has shown a paralleled improvement of anxiety and depressive symptoms as the patient's speech

becomes more fluent.[6] Currently, paroxetine and sertraline have been the only selective serotonin reuptake inhibitors that have been studied. Paroxetine has shown to be useful in the qualitative management of stuttering symptoms and theoretically reduce the cortical silent period.[25] According to Costa and Kroll,[26] sertraline has undergone placebo-controlled studies, which showed decreases in stuttering in those with persistent DS with few secondary behaviors.

Conclusion

Although there is no cure for DS, elimination of mild stuttering may be seen if intensive speech therapy is initiated early in its course. For those with persistent DS, medications can be used to decrease the rate of depressive and anxiety symptoms, which secondarily will decrease the amount of dysfluency present in speech. Therefore, general practitioners should refer patients with DS to speech-language pathologists at an early age, and those with persistent DS should have close psychiatric follow-up. At the present time, further research needs to be aimed at distinguishing the structural abnormalities present in those who stutter to discover a reliable treatment method.

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