ABSTRACT: The many theories of the cause of stuttering are too numerous to mention. Instead, this article presents a brief background of theories in the realms of psychology, learning theory, and biology. A representative sampling of older literature is discussed, followed by a more detailed consideration of a few state-of-the-art studies. The article concludes that previously presented general models can provide a basis for testable hypotheses. Current research appears to indicate that there is a genetic basis for stuttering, affecting both motor and sensory systems in the brain in subtle ways, and that deficits are exacerbated by a host of both other genetic as well as environmental factors. Genetic, physiologic, psychologic, and environmental influences guide the developmental pathway of stuttering.

KEY WORDS: stuttering, etiology, theory, speech disorder, childhood stuttering
as interesting and powerful, but many of them certainly not “true” and certainly not “scientific” to our modern way of thinking. But, given the observations that were available at the time, these theories made sense. Theoretical invention tries to suit the data. Perhaps in a millennium or two, what we now believe to be “real truth” will seem ridiculous, naïve, outdated, or wrong.

People use whatever resources they have to explain occurrences of an almost infinite number of phenomena. It has even been said that perhaps many beliefs come from the left brain trying to explain what the right brain is experiencing. And after all, truth is relative. Think of patients with various brain malfunctions. Sometimes people with aphasia have very interesting ideas about why they have difficulty with language. Children have interesting explanations for their behavior. The combination of natural curiosity and the need to explain is perhaps at the root of science.

It follows that the history of ideas about the cause and development of stuttering has undergone many developments as our knowledge base and technology have improved. It is also important to remember that science goes in circles. In other words, history repeats itself. Theories of stuttering have gone from biologic to psychologic to behaviorist and back to biologic to all three combined.

Regardless, several unusual phenomena must be incorporated into, or must not contradict, any theory of the cause of stuttering; namely, those that ameliorate stuttering. Stuttering diminishes, or even disappears, under a host of conditions—singing, speaking in unison/choral reading, talking with a metronome, talking with noise or delayed auditory feedback, and talking to pets or babies (quite similar, really, in that neither have read treatises on auditory feedback, and talking to pets or babies). Children have interesting explanations for their behavior. The combination of natural curiosity and the need to explain is perhaps at the root of science.

ASPECTS OF CAUSATION

From early on, professionals in speech-language pathology had very different views on the cause of stuttering. Stuttering has, at various points in time, been considered to be caused by deep-seated neuroses arising in infancy, purely by learning, and from a strictly structural flaw. There is general agreement today, however, that the ultimate origin of stuttering is in large part biologic, shaped by additional biologic, psychologic, and behavioral influences. We begin the review of the various aspects of causation of stuttering with a brief presentation of psychologic and learning perspectives so as to provide a full view of how far the field has come in its pursuit of truth in the name of science. This is followed by a more in-depth presentation of possible causative biologic factors. We shall not attempt to review all of the relevant literature; rather, major trends are highlighted and supported by a few samples of relevant studies.

Psychologic Causes

The writings of Sigmund Freud were in ascendance in the early decades of the 1900s. In the 1920s and 1930s, one line of thinking regarding the origins of stuttering focused on personality conflict of the individual, referencing Freud’s theory of psychoanalysis. Basically, this approach viewed stuttering as merely an overt symptom of something else—unconscious, deep-seated neurotic disorder(s). Authors purporting this point of view, however, differed in what they understood to be the specific nature of the conflict. Whereas Coriat (1928) asserted that the problem is characteristic of libido fixation at the oral stage of infant psychosexual development, Fenichel (1945) maintained that the conflict originally occurred during the anal stage, lending it more aggressive characteristics. Blanton (1931) wrote that children who began to stutter experienced fixation of the libido, thereby making the child overly sensitive and anxious. He agreed, however, that there were physiologic factors in addition to psychologic ones, such as being left-handed or having weak muscles in the tongue and throat area. Brill (1923) discussed stuttering as a psychoneurosis, although he also referred to physiologic factors as “hereditary burden” (p. 135). These authors stressed family environment as shaping stuttering. Although Brill claimed considerable treatment success using classical Freudian psychoanalysis as the main vehicle of therapy, he conceded that a large number of his former patients eventually relapsed.

The psychoanalytic point of view continued to be active, however, and in 1957, Travis wrote about “the unspeakable feelings of people” who stutter. This was a complete change from his earlier views on cerebral laterality. Travis described stuttering as a vehicle that acts to inhibit unconscious needs, or unacceptable statements of true inner thoughts and feelings. The child wants to communicate, but, being afraid of revealing unspeakable thoughts and feelings, he emits stuttered speech that conceals them. Any discomfort caused by the stuttering was, allegedly, more than overcome by the relief of hiding unmentionables. Glauber (1958) also saw stuttering as a neurotic disorder, a personality disturbance reflected in speech. Even the preponderance of stuttering in males was explained in psychologic terms in that boys faced more pressure to live up to standards of aggression and had a greater ability to provide and take action. Females, so it was said, had only to be passive creatures (Schuell, 1946).

From the 1920s through the 1980s, a large number of personality studies of stutterers were undertaken. The findings were mixed, but overall, it became clear that people who stutter (PWS) are not any more neurotic than average (see reviews by Bloodstein, 1995; Goodstein, 1958; Sheehan, 1958). The only areas identified as problematic involved communication situations, difficulties that can be explained as resulting from the stuttering rather than being a cause of it. Although today it appears perhaps illogical to consider stuttering part of a neurosis, amenable to
psychoanalytic treatment, interest in personality, especially anxiety, has been renewed, as seen in a number of recent studies. Craig, Hancock, Tran, and Craig (2003) investigated trait anxiety in a large sample of adults who stutter. Trait anxiety is a stable attribute, as opposed to state anxiety, which occurs in certain circumstances. The persons who stutter did present higher trait anxiety levels, but the anxiety appeared to have developed as a result of the stuttering and so would not be considered a causal factor. The issue is not that simple, however. As in the next study described below, it is likely that some individuals have a predisposition to develop trait anxiety, and it is not known how this tendency might interact with other factors that contribute to a predisposition to stutter.

Recently, there has been another shift of interest in psychosocial aspects of stuttering, with a focus on temperament. Anderson, Pellowski, Conture, and Kelly (2003) investigated temperament factors in young children who stutter through a norm-referenced parent questionnaire. The temperament scale used contains nine subscales: activity, adaptability, approach or withdrawal, quality of mood, intensity of reaction, distractibility, attention span, sensory threshold, and rhythmicity of physiologic functions. Compared to the normally fluent children, the stuttering children were less adaptable to novel situations, less distractible, and less regular in daily physiologic functions. The differences were statistically significantly different, but the mean scale scores for both groups on all nine scales were within normal limits. More children in the stuttering group had individual scores that exceeded 1 SD. The study also reported that there was no direct relationship between the time since onset of stuttering and scale scores, which indicates that the temperament qualities measured may be relatively stable. The point of interest to the discussion here, however, lies in a chicken and egg question: Do the differing temperament dimensions precipitate or exacerbate stuttering, or does the stuttering reinforce the temperamental proclivities of the child? The authors concluded that “temperamental characteristics...could potentially contribute, in some as yet unknown fashion, to the onset and development of stuttering” (Anderson et al., 2003, p. 1230). Although their stipulations have not yet been substantiated, the fact that there are differences means that we cannot yet rule out that possibility.

We have come a long way from thinking that neuroticism causes stuttering, but this does not mean that psychosocial factors must be relegated only to the shaping of stuttering independent of causative factors. As the genetic and environmental bases of personality reveal themselves, it may become obligatory to add them to the complex equation for the etiology of stuttering. Psychologic causes of a disorder are no longer necessarily distinct from organic causes. Overall, the renewed interest in this aspect is a welcome development. It would seem to be particularly profitable to study the interaction of personality/temperament with other factors.

**Behaviorism and Learning Theory**

The idea that stuttering is a learned behavior or a “bad habit” is quite old. Bloodstein (1995) provided an excellent overview of theories of stuttering that covers behaviorism and learning. Any discussion of behaviorism and learning theory in relation to stuttering must mention Wendell Johnson’s diagnosogenic theory (Johnson et al., 1959), stating that stuttering begins in the ears of the parents when they overreact to their child’s normal disfluencies. The child attempts to avoid the disfluencies and, in the struggle, stuttering develops. Along similar lines, Wischner (1950) portrayed stuttering as emerging in accordance with a more formal learning theory. Stuttering originates from a painful, anxiety-producing stimulation in the form of parental disapproval of normal disfluency. In other words, the child anticipates negative, painful reactions to his or her stuttering and so tries to avoid them. Initial successful avoidance reduces the anxiety drive and thus reinforces the behavior that eventually becomes more complicated stuttering.

Another concept is that stuttering results from an approach–avoidance drive (Sheehan, 1953). In this concept, the desire to communicate collides with the drive to avoid speech anxiety, which may have roots in either or both personality and conditioning from prior negative experiences with stuttering. Bloodstein’s (1958) notion that stuttering is an anticipatory struggle arising from the belief that speech is difficult contains some similar elements. Stuttering is seen as a reflection of tension and fragmentation in speech when the complexity of the act causes concern and feelings of being overwhelmed.

Still another view explains stuttering as a behavior that is acquired through operant conditioning, that is, a behavior that is shaped by its own consequences. In a pioneer, controlled experiment, Flanagan, Goldiamond, and Azrin (1958) demonstrated that stuttering was reduced when stuttered events were immediately followed by punishment (loud noise); it was increased when stuttered events were immediately followed by the removal of punishment (continuous tone). Later, Shames and Sherrick (1963) theorized that stuttering was shaped out of normal disfluency as operant behavior, subject to a combination of punishment, positive reinforcement, and negative reinforcement. Normal repetitions are reinforced when a parent eventually heeds the child’s request. But as repetitions become aversive, the parent indicates displeasure at disfluency (in terms of operant behavior, punishment). When the consequences of disfluency are negative reactions, the child may change disfluencies into struggle behavior or silence in order to avoid the aversive consequences (negative reinforcement for the parent). Thus, stuttering emerges and is maintained by complex interaction of positive and negative reinforcement. Bruten and Shoemaker (1967) developed a two-factor theory of stuttering. They believe that stuttering is initially established through classical conditioning of emotional learning, when anxiety causes disruptions in speech. Through generalization, many stimuli acquire the capability of triggering anxiety that results in fluency breakdowns—stuttering. Secondary characteristics of stuttering designed to cope with the disfluencies are then developed through operant conditioning.

In general, these ideas have been discarded as ultimate or sufficient causes of stuttering. Instead, they are now viewed
as forces in its development (see Wingate, 1997, for a thorough discussion of the matter). The idea that stuttering develops out of normal disfluencies along a continuum remains prevalent, although not supported by data, as repeatedly pointed out in the Illinois studies by Yairi, Ambrose, and colleagues (e.g., Ambrose & Yairi, 1999; Yairi & Ambrose, 1999). Nevertheless, stuttering therapies reflecting various learning models have been shown to be rather promising (Onslow, Packman, & Harrison, 2001; Ryan, 2001).

Biologic Causes

Bloodstein (1995) wrote that Aristotle believed that stuttering stemmed from a problem with the tongue, and in the 1840s, tongue reduction surgery was performed to alleviate stuttering. The better known early biologic theories of stuttering, however, come from the 1920s and 1930s. The first three years of the Journal of Speech Disorders, 1936–1938, contained no fewer than nine articles devoted to physiologic aspects of stuttering. Topics included conditioned reflexes (Moore, 1938), twinning (Berry, 1938), and heart rates (Palmer & Gillette, 1938; Travis, Tuttle, & Cowan, 1936). There are a plethora of publications regarding anatomic and physiologic bases of stuttering; only a representative few will be touched on here as a reflection of past and current work. They are grouped loosely into four sections: sensory, central processing, motor, and genetics. For each section, a brief background will be presented, and a few current studies will be discussed in more detail to indicate the general direction of cutting-edge research.

The sensory side of the picture. The role of auditory function has long been discussed in the field of stuttering. Stuttering tends to be reduced in noisy environments (as Demosthenes knew), and it may be reduced or even eliminated with delayed auditory feedback or white noise in one or both ears. This effect is even more curious in that when normally fluent speakers have delayed auditory feedback played, their speech tends to become disrupted. In 1946, Shane (1955) demonstrated that stuttering was reduced with high levels of binaurally applied noise. She concluded that when PWS cannot hear themselves, speech anxiety is reduced and so is the stuttering. A decade later, Cherry and Sayers (1956) conducted a series of experiments using both air- and bone-conducted noise of different frequency ranges to identify which one of many possible combinations is the most effective in ameliorating stuttering. Based on their results, Cherry and Sayers theorized that PWS have abnormal auditory feedback. They thought that the noise works because it neutralizes the defective auditory feedback. Therefore, they concluded, stuttering is a perceptual problem. On the other hand, Wingate (1969) suggested that noise reduces stuttering because it causes motoric alterations, such as changes in vocalization. Yairi (1976) compared monaural and binaural noise and concluded that the psychologic explanation does not hold, that the motor explanation receives some support, and that perceptual difficulties are possible factors. Hall and Jerger (1978) administered a number of auditory function tests to PWS or were normally fluent and reported subtle evidence of a central auditory problem in stuttering at the level of the brainstem.

Another method used to study the auditory system in stuttering is dichotic listening, where competing stimuli are presented to the left and right ears. Most normal right-handers show a right ear advantage for speech stimuli. Some studies showed that PWS had the expected right ear advantage, but Rosenfield and Googlass (1980) found that more stutterers than controls failed to have the expected advantage, and Blood and Blood (1984) reported that the magnitude of right ear preference was greater for those who were normally fluent. A note of caution, however, must be heeded. Dichotic listening has been described as having low test–retest reliability; in fact, in a second report, Blood and Blood (1989) reanalyzed the same data using different analysis methods and came up with varying results.

Older studies with conflicting findings have been replaced by current work with state-of-the-art technology yet again revealing a variety of differences, not always consistent, in at least some groups of speakers. Probably among the most enlightening of the newer technologies is brain imaging. Magnetic resonance imaging (MRI) can be used to create a detailed view of brain morphology, and functional MRI and positron emission tomography (PET) give an eye to function in the brain. Another promising methodology involves magnetoencephalography (MEG). Although imaging yields poor resolution in time, but excellent resolution in space, MEG is not as accurate in terms of place but gives much improved resolution in the time dimension. A few studies with findings relating to the auditory system are mentioned here.

In normal speakers, the left planum temporale, part of the auditory association cortex, is larger in the left than the right hemisphere. In some individuals who stutter, however, this structural difference is reduced (Foundas, Bollich, Corey, Hurley, & Heilman, 2001). Functional studies have also found differences in auditory association cortex (in the temporal lobe, which includes Wernicke's area) in adults who stutter as compared to those who are normally fluent. Both Braun et al. (1997) and Fox, Ingham, and colleagues (Fox et al., 1996; Ingham, Fox, Ingham, & Zamarripa, 2000) reported deactivation in this area, especially in the right hemisphere, in stuttering speakers.

One relatively recent study of particular interest used MEG to compare and contrast left versus right auditory association areas of the brain (Salmeelin et al., 1998). Participants were asked to read silently, read while mouthing but silent, read aloud individually, and read in chorus. The stuttering speakers were disfluent during reading aloud alone but fluent during choral reading. Brain activation levels (and latencies) in the right and left hemispheres in response to tones delivered to each ear during these tasks were monitored. Higher activation typically occurs opposite to the ear being stimulated, providing a contralateral response. Both groups performed similarly in terms of timing of activation (latency). But, the pattern of activation of left and right auditory association areas differed between the stuttering and the normally fluent speakers. As expected, all speakers showed higher contralateral responses.
in general, and during the speaking tasks, there was reduced activation. For all of the tasks, however, there was higher activation in the stuttering speakers, with the exception of right hemisphere activation to right ear stimuli. The second notable difference occurred in the reading aloud condition, the only task where stuttering speakers were disfluent. During this task, the balance of left hemisphere activation by the left stimulus (left ipsilateral) was decreased, and approached the level of the control group. The authors concluded that interhemispheric balance in the auditory cortex appears to be unstable in PWS.

As studies have become more and more specific and sophisticated, differences in the auditory systems of PWS have continued to be evident. All of the studies to date have been conducted with adults, and so it is not possible to separate what may be effects due to having stuttered for years from brain function as it was at the onset of stuttering. In addition, it is always important to remember that there is no evidence that stutterers form a homogeneous group, and perhaps future studies may reduce or eliminate some of the contradictions found in studies by identifying subtypes of stuttering. Any theory of stuttering, then, must take the auditory system into account.

Central processing and sensorimotor integration: A look inside the brain.

Differential hemispheric activation. In the 1930s, Travis (1931/1978) expounded on a theory of the involvement of the brain in stuttering, claiming that stuttering resulted from incomplete cerebral dominance or, more precisely, from “general reduction in cortical lead control” (p. 276). The brains of PWS have been under scrutiny continuously from that time. As technology has improved, documentation of functional differences in brain activity in normally fluent speakers versus those who stutter has become available, although results were often conflicting and thus inconclusive. For example, some EEG studies showed abnormalities in hemispheric processing in stutterers in that they used the right hemisphere more than normally fluent speakers did (Fitch & Batson, 1989; Moore & Haynes, 1980); other studies had negative results (Pinsky & McAdam, 1980). Age of participants and stimuli varied from study to study, which may explain the conflicting findings. A better explanation may be that PWS are a heterogeneous group, and not knowing how to subgroup them leads to very inconclusive findings.

Differences in hemispheric activation have been documented more recently with imaging techniques. Several independent research groups have reported greater use of the right hemisphere in speech and language in PWS as compared to normally fluent speakers (see Ingham, 2001 for a review of findings); that is to say, language is more bilaterally represented. Although an early study (Pool, Devous, Freeman, Watson, & Finitzo, 1991) reported that left blood flow in the left hemisphere was overall reduced compared to the right, areas of greatest difference were those of auditory association and motor planning.

Language. Another area within the purview of central processing is the involvement of language in stuttering. There have been investigations of phonology, syntax, semantics, and cognitive processing. It has been noted that there seems to be a higher incidence of the co-occurrence of stuttering and phonological disorders, which led to speculations about a mechanism explaining both. Postma and Kolk (1993) introduced the covert repair hypothesis, which explains not just stuttering, but all types of disfluencies, as self-repairs. Monitoring and error detection can occur at the prearticulatory level. Disfluencies are seen as by-products of covert repairs of internal speech errors, at the level of phonological encoding. When correction is successful, no error appears, but it may impede progress of an utterance, thereby leading to disfluency. Normal speakers perform covert repairs of phonological encoding. Therefore, PWS may have a deficit in phonological encoding, leading to more frequent phonological encoding errors, which must frequently be repaired, which leads to stuttering. More recent work has argued this theory (Yaruss & Conture, 1996). Other work has been directed toward linguistic complexity. As mean length of unit (MLU) and syntactic complexity increase, stuttering is more frequent (Logan & Conture, 1995; Yaruss, 1999). It has also been shown that adults who stutter may have more difficulty producing unfamiliar vocabulary fluently (Hubbard & Prins, 1994).

Bosshardt (2002) designed a study to measure the effects of cognitive interference on the speech of PWS. Two groups, normally fluent and stuttering participants, were asked to repeat sequences of three words as fast as possible (primary task) while simultaneously reading or memorizing two additional words presented visually (secondary task). The additional words were either phonologically similar or dissimilar to those of the primary task. They were then asked to recall those words. Accuracy was measured for recollection of the similar and dissimilar words that had been presented visually. For repetition of the sequence of three words, accuracy and frequency of disfluency were measured. For the control group, the concurrent reading task had no significant effect on the fluency of their repetitions, but the stuttering group had significantly more disfluencies when reading the list of similar words. In other words, their fluency-generating system was more easily perturbed by, or subject to interference from, the concurrent cognitive task. The elegant design of the study provided a way to isolate the effect of a very specific element—that of phonological similarity. This does not rule out, however, susceptibility to interference of a host of other cognitive, auditory, motor, or affective processes. Some of the brain imaging studies lend support to the hypothesis that phonological planning is aberrant in PWS, or at least in a subgroup of them. DeNil, Kroll, Kapur, and Houle (1998), and Ingham et al. (2000) reported clear differences in activation of the anterior insula in the frontal lobe, which is presumed to involve phonological planning. Another study, however, did not find such differences (Braun et al., 1997).

Sensorimotor integration. Brain activation levels can be studied using functional imaging techniques such as PET and functional MRI. These types of studies require a complex design and can be difficult to interpret. Tasks must be clearly differentiated theoretically and physiologically—a resting state, or baseline, is needed from which to measure activation or deactivation. It is not easy to determine exactly what strategies are used or what functions are
accessed or taxed when someone is instructed to view a shape versus read a word silently versus read a word aloud versus generate a synonym and so on. A newer technique yields indirect information on function through visualization of structure. Diffusion tensor imaging (DTI) allows measurement of the degree of organization of white matter, or myelin tracts, in the brain. The myelin sheaths are what neural impulses are conducted through, and the more white matter and higher organization, the better the signal. A strong signal indicates a high degree of connectivity. Areas used for comprehension, planning, and production of speech typically show a high degree of connectivity in the left hemisphere for normal speakers. Sommer, Koch, Paulus, Weiller, and Buchel (2002) studied the brains of 15 adults who stutter and 15 control speakers. They reported that the stuttering group showed significantly less fiber coherence, or myelin organization, in the area of the left rolandic operculum, which contains connections from the temporal to frontal lobes in areas subserving speech. The arcuate fasciculus, simplistically identified decades ago as the bridge between Wernicke’s and Broca’s area, is included in this area. If indeed it were such a connection that fails to perform smoothly and automatically in PWS, one would expect that the areas that it connects might show lower activation than normal. The authors further speculated that overactivation of the right hemisphere in PWS may represent a compensatory strategy.

**Motor planning and execution.** Could stuttering be due to a strictly motoric deficit? Zimmermann (1980) conducted articulatory dynamics experiments to explore this question. In the speech of adults who stutter, he described slower and asynchrony movements in their fluent speech, and aberrant positioning of articulators during stuttered speech. Zimmermann concluded that stuttering was a disorder of movement, and explained that as we speak, there is a temporal and/or spatial range of movement that is tolerable to the system. As long as the speaker remains within that range, there is no fluency breakdown. If, however, the range is exceeded, due to emotional, perceptual, and/or physiological events, the system is thrown off balance with conflicting signals to and from the brain at a reflex level, leading to oscillations (repetitions) and fixations (blocks and prolongations).

Additional studies examined the motor system as reflected in the planning, initiation, and execution stages of speech. A broad range of studies generally found slower reaction times for individuals who stutter as compared to normally fluent speakers for at least some tasks. Prosek, Montgomery, Walden, and Schwartz (1979) found differences in acoustic, but not manual or laryngeal, reaction times. Similarly, Reich, Till, and Goldsmith (1981) concluded that PWS had longer initiation times for speech vocalization, but not for nonspeech vocalization, or manual tasks. Borden (1983) had participants count out loud and count silently using their fingers and also found those who stutter to be slower, but in the execution stage, rather than the initiation stage. In Peters, Hulstijn, and Starkweather’s (1989) study of acoustic and physiological reaction times, the conditions under which normally fluent and stuttering people spoke were varied. By changing conditions, they were better able to discern the complexities of response differences. Their results indicated that reaction times for both stuttering and control groups increased with either longer utterances or minimal preparation time, but increased more for the stuttering group. They concluded that a difficulty in motor programming for speech might underlie stuttering.

Other recent studies have begun to examine aspects of reaction time. Van Lieshout, Hulstijn, and Peters (1996) tested whether or not PWS have difficulty assembling an abstract motor plan by having speakers name pictures representing one-, two-, and three-syllable words. They measured reaction time as well as duration of the spoken word. If PWS have difficulty in building a motor plan, then they should take more time for words that are longer, as compared to normally fluent speakers. They found that the stuttering speakers had slower reaction times for all of the words, but the gap between their reaction times and those of the fluent speakers remained static—that is, in both groups, performance did not appreciably change as word length grew. This evidence refutes the motor plan assembly hypothesis, and so contrasts with studies described in the previous section that reported that central processing load interferes with speech output. Overall, most studies point to generally slower laryngeal, oral, and manual reaction times for stutterers, although there are inconsistencies.

Execution of speech, as opposed to reaction times, has also been examined in PWS. Many methods have been used. Muscle activity and movement can be examined via electromyography (EMG), or second formant (F2) transition, where articulator movement is indirectly represented in the acoustic signal. Voice onset time (VOT) can be used to measure laryngeal movement. In studies of speech execution, investigators can choose to examine either stuttered or fluent speech of PWS and compare it to the speech of people who are normally fluent. Freeman and Ushijima (1978) and Shapiro (1980) found evidence of muscle activity of antagonistic pairs of laryngeal muscles during stuttering. This was thought to be at the root of moments of stuttering involving laryngeal closure. Smith, Denny, and Wood (1991), however, looked at the speech of normally fluent individuals and, surprisingly, found that they too showed activation of both adductor and abductor laryngeal muscles, negating the findings of the earlier studies. Conture, Schwartz, and Brewer (1985) also examined laryngeal behavior during stuttering using fiber optics, finding inappropriate opening and closing of vocal folds during repetitions and prolongations, indicating difficulty in motor control of the larynx during stuttering. Another interesting set of studies (Alfonso, 1991) focused on the spatial and temporal dynamics of articulator (lip, tongue, and jaw) movements in the fluent speech of PWS. Results indicated that articulatory targets were accurately reached, but articulators attained targets in a less than efficient manner in terms of the relative timing of their movements.

Webster (1988, 1989) looked at manual motor performance of PWS, their ability both to write with both hands simultaneously and to perform finger-tapping tasks. In general, he found that the participants who stutter made more errors and were slower in their performance than
normally fluent participants, and in addition they were more susceptible to interference when they were asked to perform a second manual motor task simultaneous with finger tapping. Thus, these conclusions, similar to motor speech studies, give an indication that more than just speech is involved.

Most recently, Max, Caruso, and Gracco (2003) continued to examine neuromotor skills in speech and across other motor systems. This study combined features of a number of earlier studies. Data were obtained for three motor systems—speech, orofacial nonspeech, and finger movement—and in each system, four movement sequences varying in length and complexity were tested. The use of nonspeech domains allows examination of skill levels that would not have been affected by either treatment for stuttering or years of adaptation to the disorder. If the deficit underlying stuttering extends to more than speech, then PWS would be expected to perform more poorly across the motor systems as compared to normally fluent speakers, and, as the task conditions become more difficult, their performance might be expected to deteriorate more quickly. The movements were motorically comparable and involved both flexion (closing) and extension (opening). Kinematic analyses revealed sharp contrasts in the closing versus opening movements for the stuttering versus the fluent participants. For closing movements, in the speech and finger movement tasks, the group who stutter had statistically significantly longer duration and longer peak velocity latency than the fluent group. Opening movements were not significantly different between the groups. In orofacial nonspeech movements, the same trend appeared but was not statistically significantly different. The analysis of subcomponents of the movements allowed the distinct differences to be observable without being watered down by components that were not different.

When adults who stutter are examined, it is not possible to tell if any differences found represent a deficit that caused stuttering or are due to having stuttered for years, or even are unrelated to stuttering at all. Most past investigations of young children’s speech production concluded that if differences exist between children who stutter and nonstuttering children in motor speech production, they must be subtle (see review by Conture, 1991). Some differences, however, were observed in comparisons of temporal relationships between and among related events rather than for singular speech variables (Zebrowski, Conture, & Cudahy, 1985). Recent research among very young stuttering children has provided additional indications of possible speech motor difficulties present close to stuttering onset. Hall and Yairi (1992) reported that stuttering children differed from normally fluent peers in the shimmer and jitter parameters of vocal fold vibration. Also, Hall, Amir, and Yairi (1999) found that preschool children near onset of stuttering had slower speaking rates in comparison with normally fluent controls. In fact, other investigators reported that even 1 year before stuttering onset, children’s speech contained imperceptible speech aberrations such as faster speaking rate than children who did not develop stuttering (Kloth, Janssen, Kraaimaat, & Bruten, 1995). Perhaps young children who stutter, as a group, do not exhibit pervasive deficits in speech motor function, but instead may exhibit different thresholds for perturbation.

Ideally, we would like to follow children from birth through their stuttering development, but this is extremely difficult. Another method of getting at such information is to compare traits of people who stuttered only as children with those of individuals who continue to stutter in adulthood. Traits that are the same would likely have been present close to onset of stuttering, and those that are absent in the recovered people but present in the persistent people may represent traits that normalized during recovery from stuttering. Forster and Webster (2001) used this logic to explore the idea that those who recovered from stuttering outgrew a deficit in speech motor control. (They also addressed interhemispheric anomalies, as discussed in the previous section on central processing.) The authors designed tasks to test the function of the supplementary motor area (SMA), which has been implicated as being involved in stuttering. Three groups of 24 adults participated—PWS, people who stuttered only in childhood, and people who never stuttered. There were two procedures designed to test the integrity of the SMA—a finger-tapping task and a bimanual crank turning task. The recovered group performed similarly to the control group, but the persistent group had poorer skills. The authors interpreted this to mean that for those who recovered, the SMA had matured, but the SMAs of the persistent individuals continued to function less than optimally. This type of information leads us much further into the realms of possible mechanisms not only at the root of stuttering, but also those prompting recovery.

The Fox et al. (1996) study, mentioned earlier in the section concerning central processing, found overactivation across wide areas in the right hemisphere of PWS during speaking, notably in the SMA, premotor areas, and cerebellum. When the individuals who stutter read in chorus and were fluent, many of the differences in their brain activation patterns were reduced or even disappeared. Other imaging studies have confirmed activation abnormalities in motor planning areas (see Ingham, 2001).

The motor component of stuttering clearly must be central to any theory of stuttering, as the cardinal symptom of stuttering is motoric. It would appear that PWS have a generalized motor deficit, extending to more than speech, but not perceptible in any behavior other than speech. Studies have documented slower reaction times, longer durations of motor behaviors, and effects becoming more apparent in more difficult behaviors, such as closing versus opening movements.

Genetics. Woven through some of the literature on physiologic and biologic aspects of PWS and stuttering are references to genetics (see a brief review by Yairi & Ambrose, 2002). Published research first appeared in the 1930s, when Bryngelson and Rutherford (1937) reported that the incidence of stuttering among relatives of stuttering probands is higher than in the population at large. Early studies focused on family incidence and personal characteristics, such as handedness, of PWS (Bryngelson & Rutherford, 1937). From the 1930s to 1990s, a number of
investigations (e.g., Andrews, Morris-Yates, Howie, & Martin, 1991; Felsenfeld et al., 2000; Godai, Tatarelli, & Bonanni, 1976; Howie, 1981) explored twinning and stuttering. In all, the rate of concordance for stuttering (both stuttered) was higher for monozygotic (identical) than dizygotic (fraternal twins), indicating a strong genetic factor. But, if stuttering was completely governed by genetics, then if one identical twin stuttered, his or her twin would also stutter, and that is not the case—the rate is considerably less than 100, revealing the existence of strong environmental factors.

Andrews and Harris (1964) published the first statistical analysis of the pattern of expression of stuttering in families. They found a higher risk of stuttering for relatives of females who stutter, and documented that more males than females stutter. Kidd (1984) confirmed these findings, determining that susceptibility to stuttering was transmitted vertically, that is, generation to generation, with sex-modified expression, consistent with genetically controlled traits. They concluded that males are more susceptible to stuttering, with females having a higher threshold requiring more factors for stuttering to be expressed. Ambrose, Yairi, and Cox (1993) reported different conclusions, finding that the risk to relatives of males and females who stutter is equal. Their sample, however, was very different from that of the Yale study (Kidd, 1984) in that it consisted of very young children who stutter who were followed for a period of years. Thus, the sample was representative of populations of both those who recovered and those who persisted in stuttering.

Several studies have used statistical analyses (segregation analyses) to predict what genetic model of transmission fits the observed data best. There are several models to be tested:

- nongenetic, random, or only environmental
- major contribution of one gene: single major locus (SML)
- polygenic (many genes)
- multifactorial–polygenic (MFP): several or many genes
- AND environmental factors

Andrews and Harris (1964) found that stuttering was most likely accounted for either by polygenic inheritance or by a dominant gene in conjunction with MFP components. The Yale studies by Kidd and colleagues (Cox, Kramer, & Kidd, 1984; Kidd, 1984) reported that the data were most consistent with either an MFP model or an MFP plus major locus model. More recently, Andrews et al. (1991) and Felsenfeld et al. (2000), using the Australian Twin Registry, determined that 70% of the heritability was due to additive genetic effect and 30% to nonshared environmental effect. Contributing environmental factors are those that are nonshared, or unique to the individual, rather than those that are shared by the family.

All of the studies discussed above were based almost exclusively on adults with persistent stuttering. The genetic studies conducted by the Illinois group (Ambrose, Cox, & Yairi, 1997; Ambrose, Yairi, & Cox, 1993) used a unique sample in that it is truly representative of stuttering in general by including both persistent and recovered stuttering. Ambrose et al. (1997) focused on possible subtypes of stuttering and performed separate segregation analyses for the families of children who recovered from stuttering (excluding persistent stuttering relatives) and the families of children who persisted in stuttering (excluding recovered stuttering relatives). Both analyses yielded the single highest likelihood for the presence of a major gene component (SML) in addition to MFP contributions.

Two additional findings were remarkable. First, the parameters for the SML were almost identical in each analysis, indicating that this component involves (at least) one major gene that is present in both forms of stuttering, persistent and recovered. But, for the MFP parameters, the heritability component (phenotypic variation attributed to a polygenic, additive effect) differed. Thus, the Ambrose et al. 1997 study advanced knowledge about the role of genetics a step further. The results showed that not only does the initial expression of stuttering have strong genetic components, but also that its developmental course is influenced by genetics. Specifically, the authors showed that children who stutter and who have a familial history of chronic stuttering would tend to follow that same pattern. And vice versa, children who stutter but have a familial tendency of recovered stuttering would tend to follow that pattern.

In summary, it is very likely that both forms of stuttering share a common major gene, contributions of additional genes, and environmental factors that are unique to the individual. It may be that persistent stuttering arises more from additional genetic components, whereas recovered stuttering may be less genetically controlled, with evident but less prominent family history. In addition, it is important to remember that the division of stuttering into persistent and recovered subtypes could be a false dichotomy, and that there might be a better way to classify types of stuttering. Currently, however, this appears to be the best lead.

The statistical evidence described above has laid the groundwork for linkage analysis. Linkage analysis works by identifying alleles for certain known marker genes on each chromosome that are co-inherited with the disorder in question. When a marker gene is co-inherited with stuttering, it means that the gene contributing to stuttering is on the same chromosome as the marker gene; in fact, very close to it. These regions can then be studied further to identify specific genes involved in the transmission of stuttering. Cox (2000) reported the results of the first complete standard genome-wide screen of DNA markers for analysis of stuttering performed on members of the Hutterite population in North Dakota, all descendants of a single genealogy. In this groundbreaking study, Cox mentioned sites on chromosomes 1, 13, and 17 as possible locations of genes underlying stuttering. One other linkage study has been published with weak evidence for linkage to several chromosomes (Shugart et al., 2003), and a third one by the Illinois group is currently close to completion.

**DISCUSSION AND CONCLUSIONS**

The question used to be, Is stuttering a motor disorder? A motor speech disorder? A language disorder? A genetic
disorder? A psychologic disorder? A learned behavior? Well, the answer is probably yes, to all. It all depends on what you are trying to explain. More simply put, What is stuttering a symptom of? There has been great progress in many areas, and soon researchers will begin to link up their findings. The multidisciplinary approach is absolutely necessary, although contributions can be made within any specific field. How can we put all of this information together to devise a well-formed theory? We strive to delineate the necessary and sufficient factors, differentiating cause versus contributing factor versus result. It has become clear that we cannot account for the existing data using one schema over another, but we must integrate them. Previous comprehensive frameworks, such as Smith and Kelly's (1997) multifactorial dynamic model, the demands and capacities model (Starkweather & Gottwald, 1990), or Bernstein Ratner's (1997) trade-off hypothesis, have much to offer but do not incorporate specifics as described in many of the studies discussed here. The Smith and Kelly model posits that it is the interaction of a number of components that underlies fluency breakdown, and that the contribution of each component will vary from individual to individual and over time within an individual. The demands and capacities model merely states that when demands exceed capacities, breakdown occurs, whereas the trade-off hypothesis is based on uneven resource allocation, so that if resources are diverted for a challenging task, other functions may suffer. Because these models are general, they lack explanatory relevance and are difficult to test. They were not devised to answer specifics but to provide a framework for more intricate and testable hypotheses.

We are not yet at a point to propose an encompassing detailed model of the cause of stuttering. It is safe to say that stuttering involves, at its ultimate origin, a complex but strong genetic component affecting the fluency-generating system of the brain, but this does not narrow the possibilities as much as one might think. Unfortunately, we do not yet know what such genes actually do, how they interact with each other, or how they interact with the environment. The ultimate question is, What are the types of genes that could cause the types of effects in the brain that could cause the types of aberrant brain activity that could cause the types of stuttering symptoms? Researchers in fluency are trying determine how we go from a major gene or two, some additional genes, and nonshared environmental factors to a fragile fluency-generating system that is susceptible to breakdowns related to/from factors in the spheres of linguistic complexity, time pressure, excitement or anxiety, among a host of other specifics, susceptible to interference. This mechanism must also explain how so many children stutter even severely and yet recover completely without formal intervention, whereas others stutter lifelong in spite of treatment.

Is there a specific location, or specific locations, where the process goes awry? Are all of the components in good working order, but their intercommunication does not proceed smoothly enough to allow fluent speech? Does the most primal deficit lie in auditory processing, or central processing, or speech planning? It is not easy, and probably not overly important, to determine where auditory processing becomes central processing-language-cognitive processes and where motor planning begins. All of these areas are interconnected, and a problem at one site will create a cascade effect prompting aberrations downstream. It is not as simple as finding an electrical problem in a motor, where one can systematically check the battery, the generator, the distributor, the spark plugs, and the wiring that connects them.

Although there may be, and probably are, subtypes of stuttering, there does appear to be commonality in the underlying genetically orchestrated deficit. In PWS, perhaps neural development, under genetic influence, is uneven, and systems subserving speech and language do not mature in the right order at the right time. It is known that the two hemispheres of the brain proceed differently in their maturation (Kent, 1999). The best bet may be that there is a deficit in the left hemisphere affecting both auditory and motor functions, and that the right hemisphere perhaps attempts to compensate, and one or more parts (subsystems) of the complex multilevel sensory and motor system responsible for the planning and orderly execution of fluent speech are fragile and easily perturbed. In persistent stuttering, the left hemisphere system may be wired differently and less efficiently, but may attempt to develop coping strategies, compensating with the right hemisphere. Those who stutter mildly and/or occasionally have successfully developed organized wiring mechanisms to circumvent the problem areas. Those who stutter consistently and/or severely manage to use available pathways but cannot maintain and/or develop consistent new efficient wiring. (See discussion in Yairi and Ambrose, in press). The neural system of a child who fully and naturally recovers from stuttering may develop unevenly but become indistinguishable from that of a child who is normally fluent. The challenge now is to create testable hypotheses to continue to narrow down specifics. It is an exciting time, and it is not unreasonable to expect to see exciting results around the corner.

Science has come full circle, but the spiral has tightened. In his discussion of the cerebral dominance theory, Travis (1931/1978) wrote that stuttering “reflects a certain lack of maturation of the central nervous system which either does not afford integration of the highest neurophysiologic levels involved in speech or predisposes these levels to disintegration by various types of exogenous or endogenous stimuli” (p. 277). This sounds very much like the current position on the etiology of stuttering—but now we are accumulating the concrete evidence to support it.

Relevance to Evidence-Based Practice

One’s perspective on the cause of stuttering perforce drives one’s therapeutic approach. The current thrust toward evidence-based practice demands that clinicians take a position and provide an objective rationale for their treatment regimens. This does not mean that the art of clinical judgment or client desires must be replaced by scientific findings, as Bothe (2003) pointed out. Art and science are not in opposition; rather, they should be incorporated into a unity, or consilience, of knowledge (cf.
Starkweather, 2002). Not only is the nature–nurture debate pointless and artificial, the division between researchers and clinicians is equally fruitless. As Wilson (1998) stated, in reference to science and literature, “the way to unite...is to view the boundary...not as a territorial line but as a broad and mostly unexplored terrain awaiting cooperative entry from both sides” (p. 137). Demosthenes was a philosopher and did not write a treatise on the amelioration of stuttering in the presence of noise, nor did he conduct a scientific experiment in this regard. But, his self-therapy could still be considered evidence based, using the best quality information available.

Understanding the present state of affairs of stuttering etiology theories is not a dry mental exercise, nor a mandate from both sides” (p. 137). Demosthenes was a philosopher and mostly unexplored terrain awaiting cooperative entry...not as a territorial line but as a broad reference to science and literature, “the way to unite...is to unite...not as a territorial line but as a broad and mostly unexplored terrain awaiting cooperative entry from both sides” (p. 137).

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Ambrose: Theoretical Perspectives on the Cause of Stuttering 89


