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Epidemiology of Stuttering: 21st Century Advances

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Abstract

Epidemiological advances in stuttering during the current century are reviewed within the perspectives of past knowledge. The review is organized in six sections: (a) onset (b) incidence (c) prevalence (d) developmental paths, (e) genetics and (f) subtypes. It is concluded that: (1) most of the risk for stuttering onset is over by age 5, earlier than has been previously thought, with a male-to-female ratio near onset smaller than what has been thought, (2) there are indications that the lifespan incidence in the general population may be higher than the 5% commonly cited in past work, (3) the average prevalence over the lifespan may be lower than the commonly held 1%, (4) the effects of race, ethnicity, culture, bilingualism, and socioeconomic status on the incidence/prevalence of stuttering remain uncertain, (5) longitudinal, as well as incidence and prevalence studies support high levels of natural recovery from stuttering, (6) advances in biological genetic research have brought within reach the identification of candidate genes that contribute to stuttering in the population at large, (7) subtype-differentiation has attracted growing interest, with most of the accumulated evidence supporting a distinction between persistent and recovered subtypes.

Keywords

Stuttering; Epidemiology; Incidence-prevalence; Persistency-recovery; Subtypes; Genetics

Epidemiology of Stuttering: Recent Advances

Epidemiology is the foundation of the scientific knowledge about any disease or disorder. It informs about the overall population risk of exhibiting the disorder, the risk to various sub-populations, factors that determine and influence the presence or absence of the disorder, its frequency of occurrence and various distributions (e.g., age, gender, and race), circumstance and places of occurrence, susceptibility to the disorder itself as well as to other conditions or disorders, the different courses the disorder can take, subtypes, and causes. The information

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Educational Objectives

Readers will be exposed to a summary presentation of the most recent data concerning basic epidemiological factors in stuttering. Most of these factors also pertain to children's risks for experiencing stuttering onset, as well as risks for persistency. The article also aims to increase awareness of the implications of the information to research, and professional preparation that meets the epidemiology of the disorder.

about these complex aspects is interrelated. For example, the time of stuttering onset has critical bearing on both incidence and prevalence findings (Howell, 2010; Yairi & Seery, 2011). Accuracy is compromised in incidence studies that do not extend to young enough ages because cases who exhibited early onset and natural recovery will be missed. Also due to recovery, prevalence figures vary depending upon the age group(s) examined. “Risk” can refer to the risk for stuttering onset at any point in life, that may end up in natural or treatment assisted recovery, and to the more serious risk for stuttering that persists. These two developmental paths, persistent and recovered stuttering, appear logical candidates for subtypes, but it is not yet clear what complex of biological and environmental factors determine subtypes, and indeed, there may be considerably more than two subtypes. The role of culture needs to be added to this mix. The question might be raised as to whether, in a society where stuttering is viewed very negatively, a matter of shame to be hidden, is the risk for persistent stuttering greater, or less than, a society where stuttering is more accepted?

The overall objective of this article is to highlight the scientific advances on several aspects of the general domain of stuttering epidemiology achieved during the current century, with special reference to the risk for stuttering. To this end, we examine progress pertaining to the onset of stuttering, its incidence, general prevalence, prevalence in relation to culture and other factors, developmental pathways, genetics, and subtypes of stuttering. Under each topic, background prior to the 21st century is reviewed followed by recent advances.

1. Onset of Stuttering

1a. Background

When does stuttering begin, when is the greatest risk for it to appear, and who is at risk? Studies pertinent to these questions, conducted in different countries during the 20th century, had clearly established that the great majority of stuttering cases emerged in childhood, especially its early period, sometimes even before 18 months of age (Darley, 1955; Yairi, 1983). Most studies did not report onset past age 9 (.e.g., Ohashi, 1977), including those whose samples extended to cover 22-year old persons who stutter (PWS) as in Milisen and Johnson (1936). A few cases, however, do begin stuttering during the teens (e.g., Andrews & Davis, 1964; Daskalov, 1962; Meltzer, 1934; Preus, 1981). Although there is agreement that most onsets occur in early childhood, there were substantial differences in the published data regarding (a) the mean or median age at onset, (b) the age-range in which the greatest concentration of stuttering onsets occur, and (c) the upper end of the risk window. Towards the end of the last century, Yairi (1997) calculated the mean age at onset reported in 11 studies to be 42 months (3½ years). Differences in sample size and procedures among these studies, as well as procedural flaws, however, diminish the strength of the mean just cited. Such information greatly depends on the age specifications for the sample investigated. Samples of children from birth to age 6, for example, will miss some later onsets, yielding lower mean age at onset than samples of 4 to 10-year olds that also spans a 6-year period. The latter, however, will miss the many cases of early onsets who have undergone natural recovery before age 4 (Yairi & Ambrose, 2005). Samples that use older cases will obviously result in later mean age at onset. This is illustrated in the following: Whereas in the 1983 Yairi study of children under age 4 the mean age at onset was 28 months, it was 42 months in Johnson et al.’s (1959) study of children under age 9, and climbed to 60 months in the Andrews and Harris (1964) sample of children 11- to 13-years of age.

Subjective factors may also have an impact. Inasmuch as onsets are most often reported by parents, parents of older children included in a survey present the potential problem of inaccurate memory concerning events, such as stuttering onset that occurred years in the past. For example, in their investigation of time of onset, Milisen and Johnson (1936)

questioned parents of PWS when their offspring were then aged between 3 and 22 years. The answers of some parents were based on memories of events 20 years into the past. The older the child and the longer the stuttering history, the greater the risk of faulty memories. All in all, younger samples that cover children aged between 1 and 6 years of age should yield more valid data. Stuttering onsets in adults are relatively few and mostly consequent to brain damage (see review by De Nil, Jokel, & Rochon, 2007). Some seem to be linked to emotional etiologies (Roth, Aaronson, & Davis, 1989). They have been distinguished from developmental stuttering in children because of the likelihood that they have a different etiology.

In addition to the above considerations, the question of identification arises: What do parents regard as stuttering? As reflected in several examples that follow, studies of onset convey a strikingly consistent picture. In the earliest Iowa study of stuttering onset, Taylor (1937) stated that 85% of the parents reported repetitions, especially of whole words, as the most frequent sign, 12% of the children also evidenced sound stoppages, and 11% exhibited secondary characteristics. Froeschels (1952), relied solely on parents' spontaneous comments about onset, and wrote that they specified repetitions of syllables or words as the first, and exclusive sign of stuttering in all 800 childhood cases. Also Johnson et al. (1959) reported that the majority of parents of 250 children said that the speech first labeled as "stuttering" consisted of easy repetitions of syllables, whole words, or phrases. Additionally, 12% to 15% had sound prolongations and blocks. In the Yairi (1983) study, a large majority of parents of very young children, seen close to onset, reported that the child's early stuttering consisted of syllable and word repetitions; 85% of the parents said that words (especially short ones) and syllables were repeated three to five times in a stuttering instance. A much smaller percentage of parents also listed sound prolongations, silent intervals, and blocks. In short, syllable and single-syllable word repetitions (SSW) are the prime speech characteristics that prompt identification of early stuttering by parents. Such parental descriptions have been corroborated by first-hand professional accounts. Van Riper (1982) personally examined 61 children within 3 weeks of onset and found that 80% of them repeated syllables or words; 28% had prolongations longer than 2 seconds.

The perceptual judgments discussed above have been strongly supported by investigations that analyzed recorded speech samples, such as the normative disfluency study of 90 children who stuttered, 2 to 5 years of age, all within six months of stuttering onset, and 54 normally fluent control children (Ambrose &Yairi, 1999). For the children who stuttered, Part-word Repetition was the most frequently occurring type in the class of stuttering-like disfluencies, with a mean of 5.29 per 100 syllables. Single-Syllable Word Repetition was second with a mean of 3.34 per 100 syllables, and Disrhythmic Phonation in third place with a mean 1.75 per 100 syllables. The respective figures for the normally fluent control children were: 0.56, 0.59, and 0.09. Additionally, children who stutter exhibit significantly more repeats per instance (Ambrose & Yairi, 1995; Yairi & Lewis, 1984). Based on acoustic analyses, there were also large differences in the repetition rates. The repetitions of single-syllable words, for example, were three times faster (the interval between the repeated word was shorter) for the children who stutter than for the normally fluent children (Throneburg & Yairi, 1994). Interestingly, multiple-syllable word repetitions, typically not considered stuttering, barely occurred in the speech of children near stuttering onset.

The three sources discussed above: Parent reports, clinical judgments, and speech analyses, indicate the imporance that single-syllable word repetition in early identification of stuttering. Furthermore, a very close agreement between parent and clinician's identification of stuttering has been reported by Curlee (2007) and Yairi and Ambrose (2005), the latter also showed close agreement in severity judgments.[1] Inasmuch as stuttering and normal disflueny are defined and classified on a perceptual basis that also corresponds with

quantified speech characteristics, single-syllable word repetition remains a major element in the diagnosis and definition of stuttering, just as phrase repetition is typically recognized as normal disfluency. These comments are relevant because several scholars have questioned that single-syllable word repetition is stuttering (e.g., Jiang, Lu, Peng, Zhu, & Howell, 2012; Ujihira, 2001; Wingate, 2001). In practical terms their opinion means that "an-an-an-an-an-and" is stuttering but "and-and-and-and-and-and" is not. Furthermore, one of the reasons for which Howell (2009, 2010) questioned the Reilly et al. (2009) onset findings was on account of the definition they gave to parent that included word repetition. It would be worthwhile revisiting the debate between Wingate (2001) and Yairi, Ambrose, Watkins, and Paden (2001) for an update.

1b. 21st Century Advances

Five studies with preschool age children conducted in the United Kingdom (Buck, Lees, & Cook, 2002), Denmark (Månsson, 2000; 2005), U.S.A. (Yairi & Ambrose, 2005), and Australia (Reilly, et al., 2009), have reported on the age at stuttering onset during the current century. A sixth study gathered some onset information from school children (Howell et al., 2008). Table 1 presents data on sample size and the central tendency of age at onset. As can be seen, a noticeable pattern in these studies is a lower age at onset with an approximate average of 33 months. This is 9 months younger than the mean for the 11 studies from the 20th century cited above. It is more than 6 months younger than the 41.2 months reported by Johnson et al. (1959), and more than 24 months lower than the 60 months in Andrews and Harris (1964). Furthermore, according to Yairi and Ambrose (2005), there is a large concentration, nearly 60%, of onsets in the single year between 24 and 35 months of age. Only six months later, by 42 months of age, 85% of onsets over the study range had occurred, increasing to 95% by 48 months of age. Although this particular study was open to children up to age 6, only 5% of the cases were reported to have begun stuttering past age 4. Similarly, Mansson (2000) reported only two more cases between the first screening and the last follow-up, 5 years later. That follow-up did not include a face-to-face examination but was based on the records of the five school speech clinicians who conducted the initial evaluation and continued to cover the island's entire school population. School records in Denmark are known for their completeness. In any event, our finding that 95% of the risk for stuttering onset is over by age 4 presents a substantial departure from Andrews and Harris' (1964) conclusion that 75% of the risk for stuttering is over by age 6. Although their investigation included a substantially smaller number of children who stuttered than that in the Illinois studies, it covered a longer period of time and identified several later onsets. Still, the differences in the age and the percentages are striking.

Some information has been gleaned in regard to the gender factor. In terms of age at onset, only small differences have been found between boys and girls. In all of the five preschool studies listed above, the differences were not statistically significant. That there is a significant gender bias among adults who stutter with a male-to-female ratio of 4:1 or larger has been known for many years (Bloodstein, 1995). Table 1, however, reveals considerably smaller ratios in very young children near stuttering onset; the younger the children, the smaller are the ratios.

Concerning manner of onset, there are sharp differences from traditional views and past studies. Whereas for a long period onset was described as mostly, if not exclusively, gradual (e.g., Froeschels, 1921, 1943; Johnson, et al., 1959), the recent studies reflect a strong trend for sudden onset patterns to be reported by parents. Sudden onset was reported for 40% of

[1]Other investigators also reported validity of teacher reports in which teacher diagnoses were confirmed by speech-language pathologists (e.g. Andrews & Harris, 1964; McKinnon, McLeod, & Reilly, 2007; McLeod & McKinnon, 2007).

the children by Yairi and Ambrose (2005), 50% by Reilly, et al. (2009), and for 53.2% by Buck, et al. (2002). The Howell, Davis and Williams (2008) study, conducted in the UK, had a different focus with minimal onset information obtained from considerably older children. Hence, as expected, the mean age at onset was older.

Conclusions—The recent advances show younger age at onset, similar age for the two genders, smaller M/F ratio, many cases of sudden onset sudden, and narrower age range for onset risk (a younger upper end than previously). The discrepancy seen in Howell et al. (2008) illustrates the impact of participants' age at the time of the study on the resulting data concerning age at onset. Still, such studies of later onsets may prove pertinent to several issues, for example the significance of the overlap between the typical early stuttering onset and the critical period of speech/language development (Ratner, 1997; Yairi, 1983). Is stuttering emerging after speech and language have matured different from stuttering that emerges simultaneously with the developmental processes of these domains?

Have there been real changes in the nature of stuttering onset over the past 50 years or have the improved procedures helped provide better data? If there have been changes, what is, or are, the reasons? In our opinion, the current most plausible explanation for the different findings of recent and older research is the improved procedures, especially investigators' efforts to examine stuttering as close as possible to the onset of the disorder that yield more accurate information, rather than any changes in the nature of stuttering onset over the last 50 years. The possibility that faster biological maturation as seen, for example, in earlier puberty signs documented for both girls and boys (Herman-Giddens, Steffes, et.al., 2012), also influences earlier appearance developmental disorders, such as stuttering, is intriguing but at the present also highly speculative.

The smaller polarity of affected males versus females near the time of onset as compared with the polarity at more advanced ages suggests that recovery from stuttering is considerably more frequent in girls than in boys. Data to this effect were reported by Yairi and Ambrose (1999). Thus, further investigations into the genetic basis underlying sex effects in stuttering are warranted.

2. General Incidence

2a. Background

As noted in the previous discussion regarding the time of the onset of stuttering, the incidence and prevalence of the disorder in the general population can vary greatly, depending on the time-window observed. In a narrow 24-month window between the 2nd and 4th birthday, it is quite likely that the incidence (only new cases) and prevalence (those who stutter at the time of a survey) figures will be closer than at any older ages. The reason is that as new cases emerge, they are partly balanced by existing cases who recover naturally. According to current knowledge, this particular time window encompasses the peak of both stuttering onsets and natural recovery (Yairi & Ambrose, 2005). Hence, in general, surveys that cover this age bracket provide a better indication of life-time incidence than those that begin coverage at more advance ages. Looking at a 24-month window between the 15th and 17th birthday, however, the prevalence of stuttering will exceed its incidence because, as we know, new onsets are minimal for that time-window. There are, then, several ways of investigating or analyzing incidence and prevalence. In terms of incidence, it may be reported for specific periods, averaged across several periods, or across the lifespan.

The literature on stuttering reveals a measure of confusion that has lasted for several decades regarding the meaning of *incidence*, often used when, in fact, the data reported and/or the

discussion pertain to *prevalence*. In a 1951 article published in the *Canadian Medical Association Journal*, Douglass stated that the incidence of stuttering was 1.5%. Amazingly, in a follow-up letter to the editor, Naughton (1951) referred to the above figure as "… *fantastic without basis in fact*…," claiming instead that the incidence was less than one quarter of that figure (about 0.37%). If the two authors intended to address incidence, both reported estimates were way off the currently accepted value. Furthermore, the "correction" offered by Naughton went in the wrong direction (it should be higher). More than likely, however, they had in mind the prevalence of stuttering, although even in this respect both erred, each in the opposite direction (Douglass by over-estimating and Naughton by under-estimating).

Actually, the lifespan risk for exhibiting stuttering in the population at large appears to be considerably higher than suggested by Douglass and by Naughton cited above. Bloodstein and Bernstein-Ratner (2008, p. 86–87) list 16 studies published between 1921 and 1986. Of these, 11 that had samples of approximately 1,000 or larger, reported incidence from a low of 0.7% (Culton, 1986) to a high of 15.4% (Glasner & Rosenthal, 1957). Seven studies yielded incidence exceeding 4.0%[2]. We have calculated the mean incidence for these 11 studies to be 5.88%. Still, both the sample size range from 996 to 30,586 participants, and their age that varied from close to birth to that of university students, diminish the meaningfulness of our calculated mean.

The disparity among findings is understandable because accurate stuttering incidence is difficult to determine. It can be best obtained by prospective, longitudinal tracking of large populations over a period of years that also allows direct identification and verification of all new cases which is an expensive and probably impractical ideal. The methods differ across studies and this affects results. For instance, an important factor is the participants' age at the beginning of the tracking. Initiating tracking from 12 months of age should capture *all* newly emerging cases *before* natural recovery occurs otherwise they will be lost and never counted or assessed. Starting at age 3, however, will miss some children who have already recovered; starting at 4 will miss many cases. Other factors are the frequency of case tracking: how often are children observed or re-examined, who observed them and who reported on the stuttering, and how was stuttering determined? For example, a yearly follow-up of young children is too sparse as it can easily miss children whose onset and recovery happened over a period of a few months. Additional factors are how the cases are identified, whether identification of cases is made in one, several, or many locations, whether the assessments were made by the same person or a panel, by many people who may adopt different thresholds of what they regard as stuttering, and what category of people made the assessments (parents, teachers, or speech clinicians). Also, self-identification may be different from that of other observers. Overall, cross verification using multiple samples or trained observers will result in more valid data.

The other alternative for incidence research is retrospective methods where individuals are asked to report whether they or members of their families have *ever* stuttered, that is, currently or in the past. Although retrospective studies lack the controls that are possible with more direct longitudinal methodologies, they are easier and less costly to conduct. Hence, except for two studies (Andrews & Harris,1964; Mansson, 2000) in Bloodstein and Ratner's (2008) list mentioned above, all other data were generated by questionnaire or interview-based surveys that relied on retrospective methods (e.g., Culton, 1986). Here, accuracy is compromised by informants' lapse of memory, genuine loss of information about familial stuttering history, age distribution of participants, and identification criteria. For

[2]The Seider, Gladstein, and Kidd (1983) study, reporting 13.9% incidence, was disregarded because participants were relatives of people who stuttered, a biased population.

example, a father may report that his child currently stutters. The father, however, is unaware that he, too, stuttered for several months at age 3. Such a case would have gone uncounted if (1) the father were not requested to check with his own living parents about a possible past short-term history of stuttering, or (2) had the father's parents already passed away. Another situation conducive to losing cases is over representation of young families who have yet to have more children or who have one child under age 2 who may still develop stuttering. These factors result in *under-reporting and under-counting*, yielding conservative incidence statistics. Ample examples are encountered in clinical and research settings. In our own experience with hundreds of parents, there were those who cancelled appointments because their child stopped stuttering or his/her moderate or severe stuttering improved considerably whilst they were on the waiting list for a brief period of time. Parents who brought one child who stuttered, later confided that an older sibling also used to stutter. We know parents (including two in EY's family) who simply would not think of reporting a 2-month severe stuttering episode that occurred years in the past.

Formal definition may be a factor that tightens up criteria for specifying who does and does not stutter, but its effect on incidence/prevalence studies that rely on informants' reports has not been documented. Interestingly, definitions used in two studies described below (Craig, Hancock, Tran, Craig & Peters, 2002, p. 1100; Reilly et al., 2009, p. 276), included part- or whole-word repetitions but yielded very different incidence figures for early childhood as revealed below. Many studies did not provide definitions, but rather relied on informants' perceptual judgments, (e.g. Van Borsel, Moeyaert, Rosseel, Van Loo & Van Renterghem, 2006). Relevant to this, as described in the previous section on onset, parents have judged stuttering in a remarkable similar fashion in different studies.

Concerning findings, for several decades and until the present, an approximately 5% life-span incidence seems to have been the most frequently recognized and accepted statistic. It has been cited in textbooks (e.g., Guitar, 2006; Howell, 2010; Van Riper,1982; Yairi & Seery, 2011), scientific articles (e.g., Ambrose, Cox, & Yairi, 1997; Craig, et al., 2002; Felsenfeld, Kirk, Zhu, et al., 2000; McKinnon, McLeod & Reilly, 2007), as well as in various printed and electronic sources of general information about the disorder, such as the *American Family Physician* (1998), *NIH: National Institutes of Health* (2010), *Stuttering Foundation* (2011), and others. Although similar figures appear in these and other sources, this particular estimate rests primarily on the findings of the well-known British longitudinal investigation by Andrews and Harris (1964) in which about 1000 children were tracked from the time of birth throughout the next 16 years, during which approximately 4.9% of them exhibited stuttering for some period of time. So far, this has been the only longitudinal study starting from birth and covering such a long period, and based on a population sample that some may consider as approaching an adequate size. What is adequate depends on how many children who stutter an investigator considers that the sample will yield. Andrews and Harris' 1000 children yielded only 43 children who stuttered, and the question should be raised as to whether this is sufficient for generalization? Regardless of the answer, in this study, the tracking of stuttering by health workers who made periodic home visits was rather loose and often depended on parents' reports to identify stuttering, not on direct observations by the research team (Yairi & Seery, 2011). In addition, short-term stuttering could have gone un-noticed by the examiners or un-reported by parents, making the 4.9% incidence a possible, if not likely, underestimate

2b. 21st Century Advances

Six investigations have reported incidence data since the year 2000; four of these suggest that the 5% "standard" should be reevaluated. Considering the Craig et al. (2002) survey first, it is the most extensive attempt at studying this issue. Employing a telephone interview with one member of each of 4, 689 randomly selected families (totaling 12,131members) in

New South Wales, Australia, interviewees were asked if any person residing in their home stuttered at the time or had ever stuttered (word repetition was included in the definition). The last part of the question: "…ever stuttered" makes this a potential life-time incidence study. When possible, a tape-recorded telephone interview with a family member identified by the original contact as a person who stuttered at the time, was used to confirm the case. The reported incidence, that is, the combined percent of people who stuttered at the time and those who were reported to have ever stuttered, varied from 2.8% in the 2- to 5-year old group to 3.4% in the 6- to 10-year old group. Inasmuch as the sample of 12,131 people included 263 people who ever stuttered, the life-span incidence was 2.16%. This low figure should be carefully considered in light of the second-hand information sources used (a single contact with informants over the telephone) and of all the potential sources for missing and under-reporting of cases as explained above. With respect to stuttering, it appears that underestimation is more likely than overestimation because people do not tend to claim that they had stuttered if they had not. Although stuttering is sometimes associated with cluttering, we have not encountered a "pure" cluttering case that was erroneously labeled as stuttering. (The first author did encounter one case where parents called their child's hypernasal speech "stuttering"). Recognizing these limitations, Craig et al. (2002) stated: "…this study could only provide a lower bound on the risk of stuttering" (p. 1103). Additionally, these authors did not include children under age 2 in spite of previous reported stuttering onset in this age group (see section on onset). Interestingly, only 18 children had to be discarded due to the age limit criterion. According to the Australian Statistics Bureau (2011), however, children under age 2 constitute approximately 3% of the population. Hence, there should have been about 360 children in this group, which would have probably revealed additional stuttering cases. Stuttering under age 2 has been reported by several investigators (Andrews & Harris, 1964; Darley, 1955; Dickson, 1971; Johnson, et al., 1959; Milisen & Johnson, 1936; Yairi, 1983).

The other five investigations yielded higher incidence. In Denmark, Mansson (2000) studied the incidence of stuttering on the island of Bornholm with a population of approximately 40,000. The uniqueness of this investigation is that the *entire* population of 1,040 children who were born on the island during two consecutive years was included, not just a sample. It, however, began when the children reached age 3; hence, some cases of earlier onset reported by parents to have already recovered could not be verified first-hand. On the other hand, short-term stuttering cases who had already recovered may not have been reported at all. Additionally, the child population was followed for only five years in a manner that a few additional cases could have gone undetected. Therefore, the resultant 5.09% incidence should be viewed as conservative.

Support for the increased incidence was provided the same year by Felsenfeld et al. (2000). This team investigated individuals listed with the Australian Twin Registry. There were 3,768 young adults (mean age: 23.2 years), 60.6% female and 39.4% males, who responded to mail questionnaires regarding stuttering (a small sample of PWS and controls also took part in telephone interviews). Answering the critical question if they have ever stuttered, 331 participants answered positively, amounting to 8.8% incidence. In considering this result, keep in mind that, on one hand, there is a much higher likelihood of stuttering in a monozygotic (identical) co-twin, whereas the risk for stuttering in a dizygotic (fraternal) twin is the same as for any sibling (Howie, 1981). Also, some past data hinted a somewhat greater tendency for stuttering among twins (Nelson, Hunter, & Walter, 1945). On the other hand, the substantial majority of females in the present Australian study, with a reversed F/M ratio of 1.53 females to 1 male (compared with the normal of 3 or 4 males to 1 female) would have tended to substantially lower the incidence of stuttering. Hence, there is no compelling reason to reject the 8.8% finding. Although the large percentage of females in this study may suggest that they may be more willing to respond to surveys than males, this

possibility does not affect our evaluation, that is, a sample with over-representation of females cannot be suspected as a factor that resulted in the high incidence.

Whereas the 5% and 8.8% figures are not low, one later study yielded an even higher stuttering incidence. In a 2005 publication, Månsson reported another survey conducted on the island of Bornholm. In this second round, 928 children, comprising 92% of the island's newly born children during a different set of two consecutive years, participated. Each child was individually evaluated soon after his/her 3rd birthday. The same criteria for stuttering as in the first (2000) study were employed but the procedures were more direct. Specifically, the children's speech samples were audiotaped and evaluated by the examiner to verify the presence of stuttering and to rate its severity. As in the earlier study, the children continued to be evaluated for the next 5 years. This time, Månsson and his team identified 176 children who stutter (CWS), 101 boys and 75 girls, yielding a 17.7% incidence. Whereas one is inclined to doubt such a high figure, we emphasize that, in our judgment, very careful procedures, surpassing those of the first Bornholm study, as well as other many previous studies, were employed, including diagnosis of active stuttering by both parents and two speech-language clinicians, or detailed parent reports of past stuttering which could not have occurred more than 18 months prior to the survey, and probably considerably closer. According to Mansson (2012), much of the difference between the findings of the first and second Danish studies can be explained by the greatly improved procedures. The current first author had the opportunity to observe several identification sessions conducted on Bornholm and can testify to the thoroughness of the procedures. Even if one entertains a very large 50% false positive identification, the "corrected" incidence would be close to 8%, and this covers only the first few years of life.

Next in the timeline is another twin study, this one was conducted in the United Kingdom and published by Dworzynski, Remington, Rijsdijk, Howell, and Plomin (2007). It is described in more detail in the section on developmental pathways. Based on parents' reports in a longitudinal study of nearly 14,000 twin pairs between ages 2 and 7, intermediary data available for 12, 892 children indicated the percent of "ever stuttered" was at least 8.4%. This finding bears close similarity to the 8.8% for adult twins (Felsenfeld, 2000), and is nearly identical to that of the next study described below.

The most recent and, in our opinion, methodologically most appropriate study pertaining to stuttering incidence conducted during the early part of the current century, was carried out in the state of Victoria, Australia, as a part of a longitudinal investigation of early language development (Reilly, et al., 2009). This opinion is rendered because the study was population-wide based, met criteria of young starting-age, that is, before stuttering begins, and employed professional case verification. The protocol required child health nurses to approach (consecutively) parents of all infants at their 8-month health-check visit, asking for their cooperation with the project. The original sample of 1,911 children at 8 months of age was reduced to 1619 by the age of 2 years due to attrition. It is noted, however, that the percentage (38.4%) of mothers in the participating groups who held a college degree was greater than that (29.4%) in the attrition group.

Parents were instructed to contact the investigators if/when they observed stuttering onset. A standard definition of stuttering was provided for them. In the next step, parents' diagnosis of stuttering had to be confirmed by qualified speech-language clinicians experienced with stuttering. When in doubt, a panel of two other experts viewed a videotaped sample of the child's speech in order to determine the child's stuttering status. According to this team, by age 3 incidence had already reached the level of 8.5% (137 children), a conservative count in view of the fact that 21 borderline cases (more than 13% of the total number of children, 158, referred by parents) were excluded. Had they been included, the incidence would have

climbed to 9.8%. Considering possible parent under-reporting, and expected stuttering onsets after age 3 (cases not covered in the investigators' report) as discussed earlier, higher lifespan incidence can be confidently predicted for this study.

The Reilly et al. (2009) study was criticized by Howell (2009; 2010) on two issues pertinent to its incidence portion. First, he posited that the definition of stuttering provided to parents included single-syllable word repetitions which he, and other scholars cited earlier in this context, may not, or do not, consider as stuttering. As we already discussed, backed by ample data, we disagree with this opinion. Second, Howell opined that the sound prolongation on medial vowels, specifically,"caaaaaaaaaaaaan,"[3] given to parents as an example of one sign of stuttering, is not a typical feature of stuttering. We do not have quantified data regarding the frequency of occurrence of this kind of prolongation in the speech of 2- or 3-year olds soon after onset, but we have certainly encountered them more than infrequently, if not rather commonly. Just as one example, we found the following medial vowel prolongations in a child who stutter's single sentence: "hahaaaaaaaaaaave," "claaaaaaaaaaaay, "maaaaaaaaaaaay," and "claaaaaaaaaaaay," all perceived as stuttering. Similarly, the occurrence of events such as "noooooooooooot," and "looooooooooooots," lasting 1 second or longer and perceived as stuttering, in the speech of several children who stutter, has also been confirmed by Seery (2012).

These testimonies aside, the specific example used in the Reilly et al. study ("caaaaaaaaaaaan"), appears to adequately provide parents with the idea that sound prolongations are something that they should report to the investigators as a suspected sign of stuttering. The argument that it could have confused parents to the point that, had they heard prolongations of initial sounds, e.g., "aaaaaaaaaaand," they might have failed to report it to the investigators as suspected stuttering, is not a sound one. If they did fail, then, Reilly et al.'s incidence figure is lower than it should have been. And, in case parents indeed erroneously identified stuttering based on medial vowel prolongations, their errors would have been corrected by the expert clinicians who made the final diagnosis of stuttering. This procedural detail is a crucial point. Indeed, the expert identified 21 borderline cases who were kept out of the study. Hence, Howell's reservations have little practical bearing on the Reilly et al. (2009) incidence data.

The six incidence studies published since the year 2000 are summarized in Table 2. We observe that four of the six recent incidence studies indicate a trend to up its estimate from the 5% level, with a central figure of 8% or higher. What is important to re-emphasize is that data obtained in early childhood are certainly more valid than data obtained at any other age because of the proximity of observers, both laypersons and professionals, to newly identified cases. Such data constitute *minimum estimates* because later onsets only increase the reported statistics for early life data.

It is possible, of course, that samples which supposedly represent the population at large ended up being biased, and that studies suffered procedural errors in one, or several, unknown ways. It is also possible that certain populations are different. For example, the nearly 1,000 children in the 2005 Månsson study did not constitute a sample but almost the entire population of 3-year olds on the Danish island of Bornholm at the time when the study began. In light of very strong evidence for genetic factors underlying stuttering, small, close groups with high levels of intermarriage may exhibit a higher percentage of stuttering if the disorder was present in one or several founder families. Could it be that the high incidence reflects the island's relatively low-mobility population of 40,000 that, perhaps, has

[3]The number of repeated letters designed to reflect sound prolongations is identical to that provided to parents in the Reilly et al. (2009) study

been genetically influenced over several generations by one or a few families prone to stuttering? Such a scenario, on a much smaller scale, is described in a study on the genetic of stuttering in a founder population conducted in the Hutterites in South Dakota (Wittke-Thompson, Ambrose, Yairi, Roe, Cook, Ober, & Cox (2007). Relevant information for the Bornholm study, however, is not available. Still, it seems reasonably unlikely that all the studies yielding high incidence involved samples or whole populations of very young children that were so affected.

Conclusions—In view of the above, we incline to infer that the conventionally accepted 5% lifetime incidence statistic is, perhaps, too conservative. When very careful observational and other methods are applied in research with young children, approximately 8% incidence as recently reported by several studies, high as it seems to be, should not be dismissed out of hand. We are not alone in holding this view. Bloodstein and Ratner (2008) made a similar assessment to the current one (although without the benefit of the latest information). Their review of past literature concluded that "…it would seem that a plausible figure for the lifetime incidence is *at least* as high as 10%" (P. 91). Note that they reached this conclusion without citing the Månsson's 2005 high figure, Dworszynski et al.'s (2007) 8.4 %, or the Reilly, et al.'s (2009) 8.5% findings, as the latter studies were not published at the time Bloodstein and Bernstein Ratner were writing. In any event, the new data suggesting higher incidence of stuttering have implications for other aspects of the disorder, primarily in relation to its developmental paths. These will be discussed later.

Caveats: The data presented and discussed above were derived primarily from Caucasian populations and only in North America, Europe, and Australia, although some diversity in these populations has been noted (e.g., the Australian sample reported in the Craig, et al., 2002 study). Although a few published clinical reports from several countries in different continents (see *Asha Leader*, 2005), research studies (for example, Lee & Kwak,1999, in S. Korea; Riaz, Steinberg, Ahmad, et al., 2005, in Pakistan), and prevalence surveys (e.g., Toyoda, 1959, in Japan; Okasha, Bishrey, Kamel, & Hassan,1974, in Egypt; Ardila, Bateman, & Nino,1994, in Columbia), testify to the presence of stuttering worldwide, afflicting all races, as well as many, if not all, ethnic/cultural groups, to date, no lifespan, or shorter-span, incidence study has been reported from Africa, Asia, and South America. As mentioned earlier, genetic factors may appreciably influence incidence (upward or downward) in small populations or subpopulations. Cultural forces may also contribute to incidence findings, making under-estimation particularly likely if people are reluctant to admit self or family's history of stuttering. Regardless, the similarities of prevalence data reported for these geographical areas (an issue referred to in the next section), for example, 0.82% in Japan (Toyoda, 1959) and 0.93% in Egypt (Okasha, et al. 1974) to those reported for North America, Europe, and Australia, lead us to speculate that the incidence is similar too. Ujihira (2011) also opined that specific language features can influence stuttering incidence. According to him, this is one reason why the incidence of stuttering Japan is low. Unfortunately, no recent incidence or prevalence surveys have been reported for that country to back up this contention.

3. Prevalence

3a. Background

In the disorder of stuttering, prevalence, the number of people who actively stutter when a survey is conducted, can be expected to vary with the age group sampled. As discussed above, generally, except for a short period of approximately 4 years, between ages 2 and 6, when most stuttering onsets occur, fewer cases of stuttering are expected to be identified in surveys as the age of sample increases. This statement is based on two factors. First, is the

phenomenon of natural recovery, most of which takes place within 3 to 4 years post onset, that is, before age 7 (Yairi & Ambrose, 1999; 2005). Furthermore, some recovery continues to occur during the early school grades and also beyond (Andrews & Harris, 1964; Howell, Davis & Williams, 2008; Yairi & Ambrose, 2005). Second, according to common clinical experience, as well as more systematic reports (e.g., Preus, 1981), few new onsets emerge from late childhood through adulthood.

Prevalence research is based on cross-sectional surveys, targeting a single age group, e.g., junior high school pupils, selected grades within the entire school age range, or groups according to gender, race, culture, clinical diagnoses, and others. Differences in target populations can be expected to result in a range of prevalence estimates. Unfortunately, prevalence data for discrete age groups through the entire life span within a single study of the same population pool are few. The most common procedure employed, especially with large samples, used questionnaires returned by secondary informers, such as school teachers who reported the number of stuttering children in their classes (e.g., Brady & Hall, 1976; Okalidou & Kampanaros, 2001). Direct face-to-face examination of individuals in the sample has been less common. Several surveys employed multiple procedures, such as informer input followed by face-to-face individual professional screening (e.g., Proctor, Yairi, & Duff, 2008) or checking against school records of speech-language clinicians (McKinnon, McLeod, & Reilly, 2007). Prevalence studies also have several potential weaknesses: (1) a large, diverse group of informers is likely to be deprived of a uniform perception of, or criteria for, stuttering; (2) differences in definitions may be given to informers in different studies and sometimes no such definition is provided at all; (3) samples which do not adequately represent the target groups, for example, if an investigator surveying stuttering among college age students would, unintentionally, includes a disproportionally large number of engineering majors in the sample, most of whom are likely to be males. Or, if the sample includes a large percentage of students in communication disorder programs, most of whom are females; (4) in individual screening, people who exhibit mild stuttering or show considerable fluctuation in their stuttering, may, by chance, have a “good day” and go undetected, a phenomenon we have encountered in initial evaluations as well as follow-ups.

Early studies (late 19th to early 20th centuries), conducted in geographical areas far apart from each other, involved large numbers of participants, ranging from 87,000 to 212,000, and were all limited to school children. They produced remarkably similar findings: 0.90% in Denmark (Lindberg, 1900), 1.02% in Hungary (Von Sarbo, 1901), 0.77% in Boston, U.S.A. (Hartwell, 1895), and 0.87% in six American cities (Conradi, 1904). During the next 3 decades, several surveys, mainly in the U.S.A., also yielded similar data: e.g., 0.72% (Blanton, 1916), 0.70 (Wallin, 1916). An era landmark survey of 3,471,000 children from grade 1 to grade 12, conducted on behalf of the White House Conference on Child Health and Protection (1931), resulted in an overall prevalence of approximately 0.70 % based on teachers’ reports.[4]

Studies conducted during the last third of the 20th century yielded a wider range of prevalence estimates. The lowest figure of .35% in 187,420 school children was reported by Brady and Hall (1976) while the highest was 2.12% (Gillespie & Cooper, 1973) in 5,054 junior and high school students. The findings, however, seem to center around 0.80%; for example, Leavit (1974) reported 0.84% prevalence among 10,445 grades 1 to grade 6 pupils, and Hull, Mielke, Willeford, & Timmons (1976) found 0.80% in a national sample of 39,000 grades K to 12 pupils. Although Still, Harasty and Reed’s (1994) found 1.8 %

[4]A reliability check employing a face-to-face screening of more than 10,000 pupils conducted by speech/language clinicians affirmed that finding.

prevalence for children 5–12-years old in two metropolitan Sydney (Australia) schools, it was based on a small sample of only 437 children. Our calculated mean for 9 studies published between 1973 and 1994 is 0.83%.

3b. 21st Century Advances

Several prevalence studies in specific age groups or in broad age ranges of the population have been published during the current century. Three focused on preschoolers defined here as under 6 years of age. In Greece, Okalidou and Kampanaros (2001), using teachers' responses to questionnaires, surveyed 57 kindergartens with 1113 children ages 4 to 5. Stuttering prevalence was 2.2%. In a larger study conducted in Australia (McLeod & Harrison, 2009) that also focused on children of the same age as the Greek study, findings were very different. The investigators obtained information about speech and language problems in 4,983 children, ages 4 to5 years, via parent interviews and questionnaires, teacher questionnaires, and direct assessment. Parent concern about stuttering was expressed for 5.6% of the children. A year earlier, Proctor et al. (2008) reported that among 3,165 children, ages 2 to 5, the total prevalence was 2.6%. (This study will be discussed in greater detail in the next section). Considering the younger end of the age range of this sample, the somewhat higher prevalence as compared to the Greek study makes sense.

A second Australian survey carried out by McKinnon, McLeod, and Reilly (2007) included over 10,000 children and covered a wider age-range: from kindergarten to Grade 6. In its first stage, the survey employed teacher identification. Next, the school SLPs' records were inspected in order to confirm the cases as having stuttering history. This survey yielded a prevalence of only 0.33%, a finding that stands in sharp contrast to the 1.8% reported only 13 years earlier by Harasty and Reed (1994), also for a metropolitan area in Australia. The latter, however, included only 437 children, also from kindergarten to Grade 6, although stuttering was judged by clinicians.

A large European prevalence study was carried out in Belgium by Van Borsel et al. (2006) using questionnaires distributed to teachers. It covered 21,027 pupils ages 6 to 20 with a resultant overall prevalence of 0.58 % in regular schools. Prevalence decreased systematically with age group: 0.78% for ages 6–10, 0.53% for ages 11–15, and 0.27% for ages 16–20. The M/F ratio was 4.6. In the same study, a sample of 1,272 special education school pupils, aged 6 to 15 years, the overall prevalence was higher, at 2.28%. Also covering a rather wide age-range of young people, in a much larger sample and starting at a younger age, was a study by the USA Center for Disease Control and Prevention (Boyle, Boulet, Schieve, et al., 2011). Here, researchers aimed to determine the prevalence of developmental disabilities in the general population of American youth as well as in certain sub-populations. They analyzed responses obtained between the years 1997 and 2008 from parents or legal guardians of 119,367 children, ages 3–17, who were asked if their child had any of several conditions, including stuttering/stammering, during the past 12 months. Overall stuttering prevalence was 1.60%; it was 1.99% for ages 3–10 and 1.15% for ages 11–17. Male to female ratio was 2.47.

Last to be reviewed in this group is Craig, et al.'s (2002) survey, cited earlier, that was carried out in Australia via telephone interviews with one person from each of 4,689 families, totalling 12,131 members and ranging in age from under one year to 99 years. The study is distinct for being the only one, so far, that covered the entire age range. It yielded an overall prevalence of 0.72% with variations along the discrete age groups. As predicted, the prevalence was highest in pre-schoolers and early school grades: 1.4% for children aged 2 to 5; it was 1.44% for ages 6 to 10. For ages 11 to 20 the prevalence dropped to 0.53%; for ages 21 to 50 it went up to 0.78, but down again to 0.37% for ages 51+ to 0.37%. The prevalence for ages 5 to 18 was .90%, close to the 1% level as summarized by Bloodstein

and Ratner (2008). A summary of the incidence studies reviewed above is presented in Table 3.

Conclusions—In evaluating the recent information, it is clear that prevalence under age 6 is considerably higher than in later periods in life. It means that many children stop stuttering, either naturally or with some clinical intervention, usually before that age. Natural recovery (to be discussed later) may be aided by therapy. The concentration of stuttering cases in young ages also means that preparation of clinicians to work with this age group, including evaluation, risk prognosis, parent counselling, and therapy, should be upgraded.

Three of the above cited studies that covered wider age ranges present lower, or even substantially lower, prevalence estimates than the approximate 1% central tendency noted in the past century. Note, however, that both the Van Borsel et al. (2006) and the McKinnon et al. (2007) surveys did not include children under 5 or 6 years of age, the age with the highest prevalence as was confirmed by Craig et al. (2002). Still, there seems to be a downward trend. This is especially true for the European data that, in the past, tended to be higher than 1%. Boyle et al.'s (2011) findings, however, suggest that this conclusion should be guarded. At the same time, their high prevalence figure should also be viewed with caution in view of curious data from its subpopulations to be presented later. At the present, the .72% life-span prevalence is the only one available and it also appears to be a reasonable estimate. Again, these findings, too, may have important implications for understanding the development of stuttering as will be discussed below.

3c. Race and Ethnicity

3c.1. Background—Are race and ethnicity risk factors for stuttering? Trustworthy incidence data that could provide information on the initial risk for stuttering onset as a function of race other than Caucasian, or culture, other than Western, are limited. Some past stuttering/race studies have addressed the prevalence of the disorder among African Americans. Surveys that included African American children focused on elementary school children except for Gillespie and Cooper (1973) who studied junior high and high school students. They reported that stuttering occurred more frequently in African American (AA) than in European American (EA) children. For example, 1.6% AA vs. 1.1% EA (Wallin, 1926); 7.8% AA vs. 4.11% EA (Waddle, 1934); 3.76% AA vs. 1.8% EA (Travis, Johnson, & Shover, 1937); 1.6% AA vs. 1.1% (Carson & Kanter, 1945); 2.8% AA vs. 0.7% EA (Gillespie & Cooper, 1973). The very small amount of quantified information reported from Black Africa hints at higher prevalence in certain tribes. For example, Morgenstern, (cited by Johnson, et al., 1967), cited a secondary report by another anthropologist who claimed 2.67% prevalence among 5,618 school children of the Ibo tribe in West Africa. A different picture, however, was provided by a more reliable source from South Africa. Aaron (1962) surveyed 6,581 African Bantu pupils, aged 6 to 21 years, who attended 13 schools in the Orlando township, part of the Johannesburg metropolitan area. Those who stuttered were first identified by teachers using Van Riper's definition of stuttering, and then confirmed through a personal examination by the investigator. She found 1.26% prevalence with an M/F ratio of 3.23, both figures were similar to those reported in general American and European data.

3c. 2. 21st Century Advances—Two studies reporting different outcomes, although none for overlapping ages, have appeared. Perhaps the most careful study to date pertaining to race during the 21 century so far, was published by Proctor, Yairi, and Duff (2008). It was also the only one that zeroed in on African Americans preschoolers, the age when most stuttering onsets occur. Their sample of 3,165 children, aged 2 to 5 years, comprised 2,223 African Americans and 942 European Americans. Data were collected from three sources:

individual screening by clinicians, teacher identification, and parent identification. Stuttering prevalence was 2.6% for the AA vs. 2.44% for the EA with no statistically significant group differences. Prevalence for the entire group was 2.56%. This figure is close to the 2.2% found by Okalidou and Kampanaros (2001) for 4- to 5-year olds, but somewhat higher than the 1.4% reported by Craig et al. (2002) for children of the same ages (2 to 5).

Most recently, new data have emerged from the USA Center for Disease Control and Prevention study (Boyle, et al., 2011) that surveyed parents and guardians of 119,367 children, aged 3 to 17 years, asking if their child had any of several childhood disability conditions, including stuttering. For the latter, the question specified the past 12 months as the time limit. The results reflected trends for racial and ethnic differences. For the non-Hispanic White group, prevalence was 1.27%; for the non-Hispanic Black group prevalence was more than doubled at 2.63%; for the Hispanic groups overall it was 1.96%. The difference between the White and Black groups was statistically significant at the 0.05 level of confidence. The difference between the non-Hispanic White and the Hispanic overall groups was not significant. In evaluating these data, it is important to keep in mind that much of the critical window for the onset of stuttering (under age 3) was not covered in this study. It is logical to suggest that had this age bracket been included, the overall prevalence would have risen higher. Again, the main source was parent reports with no first-hand verification even for a small part of the sample.

Conclusions: In view of the size of the Boyle et al. (2011) study, it would appear that, at present, the race factor, at least as that recognized as African American, remains opens for further careful studies. One particularly difficult question is how African-American and White are defined in light of apparent various degrees of many individuals presenting mixed races. To better examine race/genetic contribution, large, careful incidence and prevalence studies in Black Africa would be very helpful. These will also shed light on the effect of racial and cultural factors on the development of stuttering, once it has begun, in regard to persistency and natural recovery. Also the effect of ethnicity remains unclear. The racial elements in the Hispanic ethnic group are further complications that may be resolved by future research.

3d. Cultural Factors

3d.1. Background—Is the risk for stuttering culturally influenced? For several decades, especially from the 1940s to the 1960s, beliefs that cultural values can greatly influence the very incidence of stuttering were rather popular. Among others, they were influenced, on one hand, by reports from anthropologists who worked with primitive societies in New Guinea and the Pacific islands who stated that they did not encounter people who stuttered among thousands of indigenous people with whom they interacted (Bullen, 1945). These findings were reinforced by Snidecor's (1947) account that he failed to identify a single person who stuttered among 800 members of the American Indian Bannock and Shoshone tribes whom he personally interviewed. On the other hand, Lemert (1953) was under the impression that stuttering was quite common among other tribes, such as the Nootka and the Salish, a phenomenon he attributed to competiveness in their culture. Lemert (1962) provided additional examples of cultural effects on either higher (Japan) or lower (Polynesian islands) occurrence of stuttering, according to his impressions. In contrast, as mentioned earlier, Ujihira (2011) suggested that the incidence/prevalence of stuttering in Japan is low. Unfortunately, his speculations were based on personal impressions, not incidence data.

These reports were well received because they reinforced the anthropological thinking of the time as well as the prevailing diagnosogenic theory of stuttering (Johnson, 1944a, b;

Johnson et al., 1959). According to this view, the level of pressure and concern exerted by children's environment on their speaking abilities has significant consequence on stuttering. These beliefs, however, were questioned in part because of contradictory reports, such as that of Zimmermann, Liljebald, Frank, and Cleeland (1983) who found cases of stuttering in American Indian tribes earlier pronounced as stuttering-free. Furthermore, a few studies in several countries characterized by cultures that differed from the Western one, did not show markedly different prevalence from those found in American and European studies. Thus, in Egypt, a survey of 8,459 children, aged 6 to 12 year who attended either public or private schools in Cairo, yielded a prevalence estimate of 0.93 %. Stuttering was determined based on reading a standard paragraph and conversational speech. In Japan, Toyoda (1959, cited by Van Riper, 1982) surveyed 140,000 school children and reported prevalence of 0.82%. In South Africa, Aaron (1962), cited earlier, surveyed African Bantu pupils and reported a prevalence of 1.26%.

Speaking colloquially in two languages is also a cultural aspect with potential implications to stuttering risks: Could babies who are simultaneously exposed to, and learning, two or more languages spoken at home experience interference to the point that the central speech planning system malfunctions, resulting in more than the normal amount of disfluency or even stuttering? This question has implications not only for causation but also for prevention and therapy. If the answer is positive, should multilingual parents use only one language when talking to their young children or when the children are around? Should exposure to the second language be postponed until the first one is well established? Or, should special diagnostic and treatment procedures be applied for bilinguals who stutter? Indeed, the belief that stuttering is more prevalent in bilingual than in monolingual speakers has been widespread in the field of speech pathology. A review of the relevant research literature and the problems involved in such research can be found in a chapter by van Borsel (2011). Shenker's chapter (2011) is focused on the clinical aspects of stuttering in bilingualism.

Unfortunately, in relation to our current focus, past research on the prevalence of stuttering in bilingual populations has been sparse. Early on, Travis, Johnson, and Shover (1937) found that among 4,827 school children in East Chicago, Indiana, screened individually by one of the authors through listening to reading and conversational speech, stuttering prevalence was 2.8% among bilingual children compared to 1.8% in monolingual children. The difference was statistically significant at the 0.02 level of confidence. A later investigation was conducted in South Africa by Stern (1948). She surveyed 1,861 school children in Johannesburg and reported 2.16% prevalence of stuttering among bilingual as compared with 1.66% among monolinguals. The investigator also reported that three times as many bilinguals as monolinguals were judged as having severe stuttering. We are aware of no other stuttering prevalence study in bilingual populations during the 20th century.

3d.2. 21st Century Advances—No studies pertaining to the incidence/prevalence of stuttering in nonwestern cultures have been published since the year 2,000. The reasons for this lull at a time when cultural diversity has been so prominent, and when more trained speech pathologists have been working in, or in proximity to, various cultural groups are not clear to us.

Generally, little research progress has also been made in the domain of bilingualism. A procedurally problematic internet survey of 794 individuals, mostly females, from 40 countries was conducted by Au-Yeung, Howell, Charles, and Sackin (2000). Of the total participants, 656, nearly 83%, were bilingual whereas 138, approximately 17%, were monolinguals. They reported 52 different mother tongues, 70 different second languages, and an unusually high 21% life-time incidence of stuttering, especially so because the majority were females. Comparisons of incidence between the two subgroups showed no

differences with 21.65 % of of bilingual speakers reporting present or past stuttering compared with 21.74% of monolingual speakers. The lack of any control in regard to the rich mixture of those who responded, and, as suggested by Borsel (2011), the likelihood that people seek out websites of their main interests or concern cast a shadow on the findings. Such reservations were also expressed by Au-Yeung et al. (2000). Potentially important results, however, were recently reported by Howell, Davis, and Williams (2009), in England. They found that among 38 bilingual children who stuttered, ages 8–12, 95% stuttered in both languages. More significant in term of risks was the finding that children with bilingual development since birth recovered at a much lower rate than the monolingual and bilingual children who did not start to develop the second language until kindergarten.

Conclusions: The shortage of credible incidence and/or prevalence studies in different cultures during the 20th, continuing so far during the 21st century, has been a major weakness in any attempt of making the case that culture is a risk factor in stuttering. Additionally, past reports, such as those from Africa, have failed to consider separating culture from race, even in their interpretations of findings. Indeed, a major challenge to any future study in this area would be separating these two factors. Also the effect of bilingualism awaits considerably more research in order to establish basic evidence of risk. Given the size of the population involved, such efforts should be listed as one of the field's priorities.

3e. Social Factors

3e.1. Background—In regard to social-economic status (SES) as a risk factor reflected in the prevalence of stuttering, there were only two large-scale studies during the 20th century that addressed this issue. The first, reported by Schindler (1955), actually focused on educational achievements. The children who stuttered were identified in a body of more than 23,000 school children. Among other educational and psychological measures, comparisons of their parents' mean occupation level (there were 7 levels) with that of the rest of the sample did not reveal significant differences. Unfortunately, data on this measure were available for only 79 CWS. Interesting, 1/3 of the CWS were in the highest 3 SES levels as compared with only 1/6 of the nonstuttering children.

Only a year later, Morgenstern (1956) published what is by far the largest, most careful, and most influential study on this issue. He posed two questions: What strata of the population are affected by stuttering and what could possibly account for any varying incidence? To this end, 29, 299 children in the 6th and 7th grades in various size locales and several geographical areas of Scotland were screened through school personnel to identify those who stuttered who were then examined individually by experienced speech therapists to verify the diagnosis. A total of 355 children who stutter, 289 boys and 66 girls, were identified yielding a mean prevalence of 1.2%. Next, the stuttering children were distributed according to 9 classes of father's occupation, as well as 4 categories of room occupancy rate, and compared against normative distribution for 11-year olds obtained in a national study. Children who stutter whose fathers were in the class of semi-skilled weekly wage earners (e.g., machine tenders, drivers) exceeded the expected number by a greater amount than any other class. Similar findings were reported for those living in homes with fewer than two persons per room. Morgenstern conjectured that this class, which is not at the bottom of the socioeconomic ladder and is not living in poverty and despair, is characterized by the strongest desire to improve their children's future. They know that linguistic fluency is a key for progress, hence they exert more pressure and generate great anxiety when children are disfluent, transmitting it to the children. This, based on factors assumed to operate through Johnson's diagnosogenic theory, resulted in stuttering. Both lower and higher classes, where upward mobility is weaker, had lower percentages of CWS.

3e.2. 21st Century Advances—We have identified four studies, the first three of which were carried out in Australia. Keating, Keating, Turrel, and Ozanne (2001) used data from the 1995 Australian Health Survey in which information relating to the health of 12,388 children, aged 0–14 years, was collected via face-to-face interviews with primary care givers. The prevalence of communication disorders was 1.7%. SES was measured by total annual household income, career employment status, current parent occupation, and post-school education. No relationship was found between speech disorders and SES. The authors, however, did not provide a breakdown of the data according to the specific disorders. Hence, generalization to stuttering is only circumstantial. A second Australian survey of three speech disorders, including stuttering (McKinnon, McLeod, & Reilly, 2007), also found no significant relationship between prevalence of these disorders and four SES measures. It should be pointed out, however, that a child's SES ranking was determined according to the school he/she attended, not on an individual family basis.

The Reilly et al. (2009) study described earlier, also included data on the social economic status of the families involved as measured by mother's education. Among mothers of the 137 children who stuttered, 58.5% had education at the college degree level or above as compared with 36.5% among mothers of 1,482 control children who did not stutter. Statistical analyses indicated an association between maternal level of education and stuttering. The investigators stated that perhaps this result reflects under-representation of disadvantaged parents in their cohort as compared to the national population. Howell (2010) hinted that the difference might have resulted from selective attrition: the lower percent, 29.4%, of the attrition group's mothers holding a college degree as compared to 38.4% of the participating mothers. (See the previous section on onset). Our calculations, however, show that, due to the small size of the attrition group, the overall effect of the difference was rather negligible. Had attrition not occurred, the percent among the original group of 1911 mothers who held college degrees would have been 36.5% instead of 38.4%, although still lower than the national average.

In contrast to the negative findings by the first two Australian studies reviewed above, the much larger USA Center for Disease Control and Prevention survey (Boyle, et al., 2011) did find appreciable relationships between SES and the prevalence of stuttering in children aged 3 to 17 years. Furthermore, this relationship stands in sharp contradiction to the findings of third Australian study (Reilly et al., 2009). Accordingly, prevalence among children whose mothers had less than high school education was 2.57%; it declined to 1.59% for children whose mothers had high school/some college education, further declining to 0.96% for mothers who had college degree or higher. The differences between the first and third classes and between the second and third classes were statistically significant. Additionally, prevalence in children from families that were 200% under the poverty level was 2.40% versus 1.07% in children from families who were above that poverty level. Again, the difference was statistically significant. The finding that mother's education and level of poverty were associated with nearly 250% higher prevalence of stuttering, does raise questions, giving rise to doubts about the 1.60% overall prevalence reported in this study (see Table 3).

Conclusions: Until very recently, the idea that the incidence and prevalence of stuttering is influenced by the familial SES suffered from the same limitations as that of the culture factor. Although many studies of stuttering have employed some control of socioeconomic factors, the popularity of the upward mobility explanation of stuttering has been steadily declining parallel with the decline of Johnson's theory. The recent findings indicating relationships between the occurrence of stuttering and SES that have emerged from the Reilly et al. (2009) and from the large Boyle, et al. (2011) studies are contradictory as the first ties stuttering to higher SES status whereas the other associate it with lower status.

In the face of little advancement in research pertaining to incidence/prevalence in relation to culture and SES during several decades, the recently reported findings, regardless of their opposite directions, indicating that SES might be a risk factor, should revive interest in pursuing additional research into the dynamics of this possible phenomenon. The rapid progress of genetics as a major factor underlying stuttering (Kraft & Yairi, 2012), however, would seem to weaken the possibility that SES factors, if not associated with other factors, such as race, play a major etiological role in stuttering. This, however, does not mean that they do not have influences that shape the pattern of stuttering and/or its development. For example, once stuttering emerges, SES may be a factor in parents' responses to the child's stuttering and the amount of stress that it creates in the family. Whether high SES families are more stressed by a child's stuttering than low SES families is not known. It may influence parents' motivation to report stuttering, seek expert advice and/or treatment, and their interest or ability in implementing such advice. Additionally, access to other information sources, such as the internet, as well as availability of services, might be influenced. Increasing sound research in this area will improve counseling and therapy approaches.

4. Developmental Paths: Natural Recovery and Persistency

4a. Background

Once stuttering begins, the previous questions about risk change from who, and how many, will be affected to who/how many will continue with chronic, persistent stuttering, as well as who/how many will recover. Our interest here is natural recovery without clinical intervention. Information about this phenomenon has been gathered via three methods. The first is informal clinical estimates based on large clinical case loads. Johnson (1934) and Bryngelson (1938; 1943) were among the first to report approximately 40% recovery without treatment by age 8. The latter's observations were based on a 1,492-strong caseload.

The second method employs retrospective techniques. One way has been through asking parents about the speech and stuttering history of their children. For example, Glasner and Rosenthal (1967) reported 54% recovery while the Dickson (1971) study yielded 58% recovery. Other investigators, e.g., Sheehan and Martin (1966), asked university students about their own present or past stuttering history. They found an 80% recovery rate. Such studies, however, lacked speech-based data to verify the initial stuttering diagnosis as well as the later remission. Reviewing 15 reports, all except one in these two categories, Wingate (1976) calculated the overall recovery rate at 43% by age 14 but thought that the recovery rate may be well above that. We have already emphasized that when adults report about themselves, or when parents report children's stuttering far in the past, memory lapse and lost information results in underestimates.

The third approach uses longitudinal strategies that monitor children's stuttering over several or many years. If tracking begins at a young age, close to the time of onset, if follow-ups are sufficiently frequent, and the number of children who stutter is large, this method provides more accurate data. The first study of this kind was the 16-year longitudinal investigation that began at birth was carried out in Britain by Andrews and Harris (1964). It yielded 79% recovery without treatment and brought the phenomenon to the forefront. An example of a problematic study was conducted in Sweden by Fritzell (1976). With the initial visit /examination taking place between ages 7 and 9, it was much too late to capture a large percentage of the recovery that typically occurs in earlier years. They found only 47% natural recovery.

During the last 3 decades of the 20th century, several longitudinal studies were published. In the USA, Panelli, McFarlane, and Shipley (1978) reported 80% recovery rate. Yairi and

Ambrose (1992) reported the first systematic speech-based follow-up study (2 or 3 times per year) of early childhood stuttering that began soon after stuttering onset. This small pilot study of 27 children that tracked the frequency of stuttering over time indicated 67% recovery after two years, growing to 89% a few years later. In 1999, these two authors reported 74% recovery after 4 years for a much larger n of 84 children. Also toward the end of the 20th century, Kloth, Kraaima, Janssen, and Brutten (1999) in the Netherlands found 70% recovery while in Germany, Rommel, Hage, Kalehne, and Johannsen (1999) found 71% recovery.

4b. 21st Century Advances

Recent research in several countries in three continents has consistently yielded a high rate of natural recovery, ranging from 68% to 96%, when children began participation during preschool years. Of the 9 studies listed here, 7 employed young children and two researched adults. Eight studies pursued the longitudinal methodology and one was based on retrospective data. Longitudinal investigations in Denmark yielded 85% and 94% recovery (Mansson, 2000 and 2005 respectively). These two studies are unique, as mentioned, in that each included nearly all children (close to 1,000) who were born during two consecutive years on the island of Bornholm, hence, they were not selected samples. In Germany, Johannsen (2001) reported 77.4% recovery by the 9th sixth month follow-up after 4.5 years into the study. These were updated results of the initial report by Rommel et al. (1999) mentioned above. In the United States, recovery was reported at the 68% level (Ryan, 2001), and 79% (Yairi & Ambrose, 2005). The latter presented updated information, over a longer time period than Yairi and Ambrose (1999).

By far, the largest investigation that provided information concerning persistency and recovery has been a part of the Twin's Early Development Study (TEDS) conducted by Dworzynski, Remington, Rijsdijk, Howell, and Plomin (2007) in the United Kingdom. The project included parental questionnaire responses of close to 14,000 pair of twins who initially agreed to participate. At each of four ages (2, 3, 4, and 7), a part of a questionnaire distributed to parents asked them to comment about stuttering in their children (among many other health and developmental items). Not all parents, however, responded at all of the four stages. The last follow-up that the authors included in the article, at age 7, covered 12, 892 children for whom at least two ratings of stuttering were available. Within this group, children for whom stuttering was positively indicated at one or more earlier ages and were still stuttering at age 7, were designated as *persistent*. Children for whom stuttering was positively indicated at one of the earlier years but was not indicated at the last follow-up, were designated as *recovered*.

The results showed that, by age 7, 1085 of the 12,892 children were marked for stuttering. Of these, 950 (87.55%) recovered and 135 (11.39%) persisted. This recovery rate is close to Mansson's (2000) 85%. Also, Yairi and Ambrose (2005) opined that, due to a good number of initially underreported cases they encountered, the real rate of recovery is higher than the 75% that they found. (In the opinion of the authors, it is close to 90%). Keep in mind, however, that the Dworzynski et al. findings do no not include stuttering verification by objective speech data, nor were they informed about possible speech therapy.

Two longitudinal studies, limited in follow-ups, were carried out in the United Kingdom with older children. Howell, Davis and Williams (2008) had 76 children, 64 boys and 12 girls, ages 8 to 12 who had stuttered for a while and received a brief therapy program. They were evaluated upon entering the study and data were only reported for when they reached age 12 (data at other ages are available for subsequent analysis). With 50% recovery rate, the investigators showed recovery to continue during the school years, though at a lower

rate. In a repeat investigation of 206 CWS, Howell and Davis (2011) found approximately 52% recovery after 6 years on the average.

Finally, retrospective data were included in the incidence study by Craig, et al. (2002), which surveyed a large sample of 4,689 families with more than 12,000 members, covering the entire age range and representative of the population of the Australian state of New South Wales. This team reported 70% recovery over lifetime. Table 4 summarizes these studies.

In addition to the three approaches to estimating recovery, it is also possible to apply a fourth method, that of statistical deduction. This indirect, but potentially powerful method, uses the disparity between the population-wide incidence and its corresponding prevalence. Conservatively speaking, if the lifetime incidence of stuttering is the classic 5%, and the average prevalence is the classic 1%, it can be deduced that the persistency rate is 20% (1/5) and the recovery rate is 80% (4/5). The advantage of this calculation is its large body of data derived from various studies that, combined, have included millions of individuals screened for stuttering. Although different methods have been used, there is power to the very large numbers and their overall pattern.

Conclusions—Recent studies on natural recovery in diverse geographical areas support the early report of high recovery rate by Andrews and Harris (1964). Natural recovery findings have inspired some controversy (e.g., Ingham & Bothe, 2001) and one may expect different results in new studies. Nevertheless, the large population-wide incidence-prevalence disparity is indisputable and stands out as a strong evidence for natural recovery. The recently reported high incidence, for example 8.4% by Dworzynski et al., (2007) and 8.5% by Reilly et al. (2009), has significant implications for natural recovery. Using the last figure and the traditional 1% prevalence, the persistency rate is 11.7% (1/8.5) and recovery rate is 88.3% (7.5/8.5). If the prevalence is lower than 1%, as some recent studies suggest (e.g., 0.72% by Craig, et al., 2002, and 0.58% by Van Borsel, et al., 2006), and incidence is about 8%, recovery is even greater, a whopping 91%. Interestingly, on the one hand recent incidence data suggest a *greater* initial risk for exhibiting stuttering than what has been thought for a long time. On the other hand, comparisons with the updated prevalence data indicate that the percent of risks for persistency is *lower* than has been suggested. In other words, the long term prognosis is more favorable. In fact, longitudinal studies have served to confirm the high positive prognosis as deduced from data on the population at large. Although some participants in incidence/prevalence studies who recovered also received therapy, most likely their numbers were small within the large-scaled surveys conducted in the past, when clinical services for stuttering were sparse. Incidence and prevalence studies in other countries where clinical services are absent remain sparse. Once these become available, they could provide additional desired information. Longitudinal studies are particularly useful for the purpose of identifying criteria for risk prediction of persistent stuttering in early childhood (Yairi & Ambrose, 20005) as well as in late childhood (Howell & Davis, 2011).

5. Genetics

5. 1. Background

Research concerned with the possible contribution of genetics to stuttering began with the *family incidence* method, counting the percent of PWS having relatives who stutter (e.g., Wepman, 1939; Poulos &Webster, 1991). It was rather inaccurate and could easily be misleading because the family sizes of the probands or the familial class of the stuttering relatives, for example, were not considered. Nevertheless, the accumulated large body of data, showed a large presence of familial stuttering, encouraged more refined research, using

the method of *twin studies*. Investigations reporting that the percent of concordance for stuttering among identical twins who share identical genes, was higher than in non-identical twins (e.g., Godai, et al. 1976; Howie,1981). This lent stronger support to genetics' role in stuttering. The next, more advanced level, employed family aggregation studies that accounted for the probands' pedigrees. The detailed distribution of stuttering among classes of relatives: mothers, fathers, brothers, sisters, grandparents, uncles, aunts, and cousins, allowed for the assessment of their respective risk. Application of the statistical technique of *segregation analysis*, yielded essential information of whether or not, and how well, the data fit with existing transmission models, such as single major locus, multifactorial polygenic, and a mix of the two (e.g., Ambrose, Cox, & Yairi, 1997; Cox, Kramer, & Kid, 1984; Kidd, 1980). The good match with several models provided additional important support to the genetic bases of stuttering. A comprehensive review of the relevant literature may be found in a recent publication by Kraft & Yairi (2012).

5. 2. 21st Century Advances

The current century has seen a surge in twin studies conducted in three continents. Felsenfeld et al. (2000) surveyed 1,567 pairs and 634 singles located in Australia to identify those with stuttering history. They concluded that approximately 70% of the variance in liability to stuttering was attributable to additive genetic effects, the remainder being due to nonshared environmental effects. In Japan, Ooki (2005) investigated a similar sized sample of 1,896 twin pairs. Concordance for stuttering in monozygotic pairs was 52% but only 12% in dizygotic pairs. The genetic contribution to stuttering was estimated at 80% for males and 85% for females. Most recently, Beijsterveldt, Felsenfeld, and Boomsma (2010), in the Netherlands, looked at 10,500 five-year-old Dutch twin pairs. Through a questionnaire, parents were asked about frequency of repetitions, prolongations, and blocks observed in their children. Responses were categorized as "probable stuttering" or "high non-fluency." Concordance for probable stuttering across genders was 57% in monozygotic vs. 31% for dizygotic pairs. These results are in concert with the findings generated by other twin studies that support stronger phenotype inheritance in identical twins.

The large twin study conducted by Dworzynski, et al. (2007) in the UK described earlier, had one focus on the phenomena of recovery/persistency (950 CWS recovered and 150 persisted). Concordance rates were consistently higher for monozygotic than dizygotic twin pairs. Using liability threshold modeling, they determined that stuttering beginning in early childhood is highly heritable, with significant additive genetic and nonshared environmental components (with little shared environmental effect), confirming prior reports by Ambrose, et al., 1997 and the several studies mentioned immediately above. The additive genetic-nonshared environmental model was the best fit for both persistent and recovered stuttering. Whereas Ambrose et al. (1997) reported differences in the genetic components of the two groups, the British team found no difference in the best-fitting genetic model for persistent vs. recovered stuttering.

Other contributions were in the work of Viswanath, Lee, and Chakraborty (2004) who, through segregation analysis of data derived from 56 pedigrees of PWS, showed an autosomal dominant major gene effect in stuttering influenced by the sex and affected status of parents. (A major gene involvement was first shown to be statistically significant by Ambrose, Yairi, & Cox, 1993). Finally, Yairi and Ambrose (2005) affirmed their earlier findings, reporting that whereas 88% of children who persisted had positive familial history of stuttering, only 65% of children who recovered naturally had such history (n = 123).

The strong evidence for major genetic components to stuttering accumulated during the 20th century through statistical genetics, reinforced by studies conducted during the current century, paved the way for the next step, employing biological genetics methods to analyze

DNA materials with the aim of locating and understanding candidate genes that underlie the disorder. Four methods have been pursued, yielding 8 studies. The methods and their respective studies are presented below in chronological order.

The linkage analysis approach relies on genetic markers to identify variations within families. Markers are DNA segments that have been previously identified for their chromosomal locations and properties; hence, they can serve as nautical points of reference. The technique is based on what is known as co-inheritance, or linkage, where genes that are physically close to each other tend to be inherited together. Genes close to a marker are genetically linked with it and will be co-inherited. Linkage analysis identifies DNA segments that are close to markers and are inherited by family members affected by a disorder, stuttering in our case, but *not* by unaffected members. This, then, provides clues as to the possible location of the target genes. Four studies are listed below[5]

1. Shugart, Mundorff, Kilshaw, Doheny, Doan et al. (2004)*. Data were obtained from 68 North American and European families, each with several people who stutter. Of the 226 members, 188 stuttered. A low-to-moderate linkage to stuttering was found on chromosome 18. Weaker linkage signals were detected on chromosomes 1, 2, 10, and 13.
2. Riaz, Steinberg, Ahmad, Pluzhnikov, Riazuddin, et al. (2005)*. Used data from 44 PWS and 55 non-stuttering members of 46 highly inbred Pakistani families. The best linkage was found on chromosome 12, with weaker signals on chromosomes 1, 5, and 7. Questions are raised, however, because with inbred families the findings may apply only to their specific genetics.
3. Suresh, Ambrose, Roe, Pluzhnikov, Wittke-Thompson, C-Y Ng, et al. (2006)**. DNA was extracted from 100 American, Swedish, and Israeli families with 252 people exhibiting persistent stuttering, 45 recovered from stuttering, and 19 who stuttered but were too young to be classified in one of the two groups. A moderate evidence for linkage for all 316 participants who ever-stuttered was detected on chromosome 9. Weaker signals were located on chromosome 15 for persistent stuttering and on chromosome 2 for natural recovery. The strongest evidence for linkage in the study was for females only on chromosome 21. A signal for males only was found on chromosome 7. Findings indicated that stuttering may result from the contribution of several chromosomal combinations, such as 9 and 2, or 7 and 12.
4. Wittke-Thompson, Ambrose, Yairi, Roe, Ober, & Cox (2007)**. This study was carried out on a founder population that encompassed a 232- member Hutterite family from South Dakota that included 48 PWS. A genome-wide mapping yielded nominal evidence for linkage with stuttering on chromosomes 3, 9, and 13. Combining the Hutterite samples with the 100 families used in the third study above, additional nominal evidence for linkage were found on chromosome 2 and 5.

   Studies #5 and #6 employed candidate gene analysis:

   A more advance stage is *candidate gene analysis* that limits exploration to smaller, but more focused, DNA locations, looking for specific genes already suspected of being involved. Such narrowing is based on knowledge concerning the biological

[5]* denotes NIH team headed by D. Drayna; ** denotes Illinois International Stuttering Research Program team, headed for these studies by N. Cox, Univ. of Chicago, Nicoline Ambrose and E. Yairi, Univ. of Illinois.

characteristics of the disease, sufficient to intelligently propose specific candidate genes. The two available studies are described below:

5. Lan, Song, Pan, Zhuang,Wang, et al. (2009). This team obtained DNA from 112 Chinese who stuttered and from matched controls. Based on previous reports that a good number of children who stutter also suffer ADHD problems, they searched specific regions that contained two genes, SLC6A3 on chromosome 5, and DRD2 on chromosome 11, known to be associated with ADHD. Both involve dopamine control in the brain. Only one significant variation within the DRD2 gene was found, supporting the notion that dopamine excess involved in stuttering. The basic tenant of high frequency of ADHD among people who stutter, however, is questionable.

6. Kang, Riazuddin, Mundorff, Krasnewich, Friedman, et al. (2010)*. Following several rounds of analysis that focused on a single highly inbred Pakistani family, this team identified a mutation in the GNPTAB gene (on chromosome 12) in most of the stuttering members. The gene has a metabolic function. The team identified the same mutation in five other stuttering individuals from their Pakistani population as well as in one North American person of Asian-Indian ancestry. Additionally, three mutations of another gene, NAGPA on chromosome 12, were identified in 6 affected North American-British participants but not in the Pakistani population. These genes function in relation enzymes that direct lysosomal metabolism in the brain and body. Although these were initially celebrated as the first discovered "stuttering genes," the Kang, et al. study was criticized by several scholars (Fisher, 2010; Büchel & Watkins, 2010) and was severely evaluated by Kraft and Yairi (2012) stating that "…the study offers little information that can be generalized to the population at large of people who stutter."

The third, approach is *genome-wide association study* (GWAS). It is used to associate specific genetic variations with particular diseases. The method involves scanning the genomes from a large number of unrelated people, looking for genetic markers that can be used to predict the presence of a disease. Once such genetic markers are identified, they can be used to understand how genes contribute to the disease. One recent investigation employed this method:

7. Kraft, SJ., (2010)**. The investigators used DNA from 88 Americans and Europeans people who stutter, aged 13 to 70 years, and from 107 nonstuttering controls. Candidate genes were selected via identification of regions that demonstrated allele frequency differences between the stuttering and the control groups, and from those identified in previous studies. The author's preliminary report indicated 10 candidate genes to be significantly associated with persistent stuttering. Furthermore, all are critical to many neurological processes related to speech production. Of these, 8 are associated with known neurological and/or motor disorders. Findings of this study should be viewed with caution as they have not yet been scrutinized through peer review.

The fourth and last approach is functional genome-wide association (fGWAS). Whereas GWAS looks across the genome and does not rely on relatedness of participants to others participants, fGWAS probes via statistical procedures into which genes are interacting with each other and reveals underlying pathologies to the disorder. The most recent investigation employed this technique:

8. Kraft, Cox, Evans, Ambrose, & Yairi (2012)**. In this preliminary report, the investigators presented an extension of the research described in study 7 above. The results of fGWAS suggest that the pathology underlying stuttering may involve

zinc and/or glucose mitigated neurological functions. Again, findings of this study should be viewed with caution as they have not yet undergone peer review.

Conclusions—As we have gradually moved from the crude method of family incidence into the most advanced genotyping techniques, evidence for a strong genetics factor in stuttering has become unquestionable. Although it is too soon to declare that specific genes underlying stuttering in the population at large have been isolated, it appears that the few scientists in the forefront of this research have come close to identifying a number such genes, perhaps even gaining some understanding of how some of them function. Replications and confirmation of recent initial reports, however, are a must. Finally, because multiple genes appear to underlie stuttering, understanding of their complex interaction remains difficult.

6. Stuttering Subtypes

6.1. Background

The rich diversity of overt and covert stuttering symptomatology invited various subtype classifications proposals during the 20th century. They tended to involve a single dimension and were accompanied by either little or no supporting research-based data. For example, etiological based classifications: Brill's (1921) psychogenic subtypes and Canter's (1971) neurogenic subtypes. A symptomatology-based classification, e.g., tonic vs. clonic disfluency, was offered by Froeschels' (1943), which is also reflected in Schwartz and Conture (1988). A biological characteristics classification was proposed by Seider, Gladstien, and Kidd (1983) based on positive-negative family history. J. Riley (1971) proposed a differentiation system that rests on concomitant conditions: children exhibiting concomitant language deficits only, and those exhibiting inferior motor skills.

In spite of the many subtype schemes that have been floating around (see reviews by Seery, Watkins, Mangelsdorf, & Shigeto, 2007; Yairi, 2007), little research has been done and no current stuttering theory accounts for subtypes. One example of such research is seen in Kroll's (1976) study. He apriorily classified adults as exhibiting interiorized or exteriorized stuttering, then compared them on the Rotter Locus of Control Test, recorded speech samples, and several psycho-social and speech variables. Findings indicated that severity of stuttering, concern about it, and group affiliation, identified a large majority in each group. Hinkle (1971) studied the lateralization of dichotic listening in adults who stutter as an indication of brain asymmetry in auditory processing. Three subgroups emerged: right ear preference, left ear preference, and no preference. The subgroups also differed appreciably in their stuttering patterns and severity, as well as in the level of the adaptation effect (decrease in overt stuttering on repeats of the same material).

A few studies were conducted close to the end of the 20th century. Schwartz and Conture (1988) expanded on the idea of contrasting tonic and clonic disfluencies in subtyping. They observed 43 children who stutter, aged 3–9 years, and identified subgroups by analyzing the type of disfluencies (repetitions and sound prolongations) and 14 other speech and nonspeech behaviors. Cluster analysis of the children's scores revealed five subgroups which the investigators reduced to just two: stuttering children who predominantly exhibited repetitions (clonic), and those who predominantly exhibited sound prolongations (tonic).

In a large study, Poulos and Webster (1991) examined 171 people who stutter, 112 with positive familial histories of stuttering and 57 with negative histories. Only 2.4% of the first group reported birth or developmental trauma or illness as compared with 37% in the second group. The findings supported two subtypes: one having a genetic basis and a second in

whom stuttering was induced by some form of prenatal or early childhood physical trauma that perhaps caused brain dysfunction.

Ambrose, Cox, and Yairi (1997), investigated subtypes in respect to diverse developmental paths or tracks, i.e., natural recovery and persistency, looking for genetic bases. They performed segregation analyses on the pedigrees of 66 young children who stutter to examine patterns of heritability. The analyses provided statistical evidence for both a single major locus and polygenic component for persistent and recovered stuttering. The data, however, suggested that persistence is, in part, due to additional polygenic components not seen in children who recovered naturally.

6.2. 21st Century Advances

Encouraging research activity has taken place during the current century. First in the time-line was a study that again pursued subtype classification based on overt disfluent characteristics. Fienberg and Levy (2000) compared 12 tonic and 18 clonic adults who stuttered on several tests. They found that the two groups could be discriminated along personality, cognitive, and intellectual dimensions. No additional research pertinent to this classification has been reported since then.

A completely different direction has focused on brain morphology and function. In their study of 14 adults who stutter, Foundas et al. (2004) reported that a subgroup of 5 participants with atypical rightward brain asymmetry, that is, atypically large right planum temporale (a section in the Wernicke's area that is important for auditory processing) was significantly more disfluent at baseline condition than were the 9 subjects with typical PT anatomy (leftward asymmetry). The two groups also differed in their response to altered auditory feedback (AAF). PT asymmetry appeared to also be associated with the effect of auditory feedback on the frequency of stuttering. Whereas the right asymmetry subgroup became fluent when speaking under DAF, the left asymmetry subgroup did not. The results indicated that anomalous anatomy of the PT, may, in this case, be associated with aberrant auditory processing. These findings suggest biologically-based subgroups. One should note, however, that 5 of the control participants also had rightward planum temporale asymmetry, showing this condition is not specific to stuttering.

Still another direction that has enjoyed considerable more research in recent years has re-focused on stuttering developmental paths, that is, persistent and naturally recovered stuttering. The question has been about whether individuals who follow these paths are also distinct in other respects. Data have been accumulated separately from several domains. Forster and Webster (2001) postulated that stuttering is associated with anomalies of interhemispheric relations and of the neural mechanisms of speech-motor control, specifically those involving the supplementary motor area (SMA). Accordingly, natural recovery is related to a maturation of the SMA. They reported differences between 24 adults who persisted in, and 24 who recovered from, stuttering on two motor tasks, finger sequencing and bimanual crank turning. The recovered group performed similar to the controls but the persistent group had poorer skills. The investigators concluded that recovery from childhood stuttering reflects a maturation of the mechanisms of speech-motor control. This, to us, is a rather unspecific term.

Supporting brain morphology data were provided by Chang, Erickson, Ambrose, Hasegawa-Johnson, and Ludlow (2005). Of 14 boys who stutter, aged 9 to 12 years, 7 exhibited persistent developmental stuttering and 7 had naturally recovered from stuttering. MRI scans were used to assess regional gray matter density (GMD). Children who recovered had less GMD in bilateral cerebellar regions compared to either the persistent or the control group. The recovered group also exhibited significantly *less* GMD than the persistent group in the

left medial temporal gyrus/superior temporal gyrus, right inferior parietal lobule, and bilateral precentral gyri. A year later, genoytyping of DNA extracted from blood samples of 316 individuals who stuttered also indicated genetic underlining of the persistent-recovery subtypes. Suresh, Ambrose, Roe, et al. (2006) reported that whereas the strongest gene signal for people who stutter as a whole was located on Chromosome 9, the strongest signal for people with persistent stuttering was detected on Chromosome 15. Most intriguing to the subject of subtyping was the indication that genes from three different combinations of chromosomes (#2 and #9; #7 and #12; #7 and #18) may result in stuttering. Finally, the distinction between persistent and recovered subtypes took an additional forward step into clinical applications. Summarizing the Illinois Studies on the development of early childhood stuttering through longitudinal follow-ups, Yairi and Ambrose (2005) reported several early (soon after onset) predictors for the two tracks. These include specific patterns of family stuttering history, gender, age at onset, and several measures of stuttering patterns.

Whereas past ideas were limited to a single dimension of the disorder, approaching stuttering subtypes from several domains simultaneously is promising to be more productive. Initial data assembled in a large research project, of which pilot findings were just reported (Ambrose, Yairi, & Loucks, 2012) include information obtained from over 200 longitudinal (for preschool) and cross-sectional (for school age and adults) participants who stutter, as well as control subjects and parents, totaling 851 participants. The possible emergence of subtypes is being investigated by integrating data from four domains: epidemiology, motor, language, and personality, applying multivariate techniques that parcel data into clusters.

Conclusions—Whereas past interest in stuttering subtypes did not produced reliable and clinically practical results, the recent rise of interest in this area, seen in the few studies just reviewed and in the central importance of the topic in the program of the 2012 World Congress on Fluency Disorders, is encouraging. We note that the persistent-recovered subtypes have received the greatest research attention during the current century and that positive findings were reported. Most of the recent studies, however, were too small in scope (e.g., 14 participants in each of the Foundas et al., 2004, and the Chang, et al., 2005, studies) and should be viewed with due caution. Larger in scope investigations integrating multiple domains, should be more promising.

General Summary

There is a burgeoning body of knowledge of epidemiology of stuttering with reference to the six areas discussed in this article. Whereas the early part of the 21st century has seen significant new information in all six areas, most consequential have been the advances in genetics and, potentially, the diverse developmental paths of the disorder during early childhood. Recent data on incidence and prevalence also reinforce findings regarding the large numerical disparity between these paths: Recovery and persistency. With further advances in these areas, and with additional contributions from genetics, the 21st century may well witness, within a decade or two, very significant progress in our ability to predict risk of persistent stuttering. Data on the incidence and high prevalence of stuttering in preschool age children call for upgraded preparations of clinicians for working with this age group, including initial evaluation and prognosis of risk for persistency, parent counselling, as well as for treatment of children considered to be at risk. In spite of the fact that sooner or later many children experience natural recovery, their initial large number also requires greater availability of clinical services. Technological advances in genetic analyses and brain imaging, coupled with improved data from large long-term behavioral studies, are yielding rich information on onset, incidence, prevalence, developmental pathways, and

subtypes. It is through the integration of a wide spectrum of studies that we can better understand the disorder of stuttering.

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Biographies

Ehud Yairi (B.A., Tel Aviv University; M.A., Ph.D., University of Iowa), has contributed extensively to the research literature on stuttering with a special focus on the various aspects of the onset, development, and genetics of stuttering. He authored many scientific articles as well as two books: Early Childhood Stuttering (2005; with N. Ambrose), and Stuttering: Foundations and Clinical Applications (2010; with C. Seery).

Nicoline G. Ambrose received her PhD from the University of Illinois. Her research centers on the etiology, onset and early development of stuttering, with particular reference to genetic factors underlying possible subtypes of stuttering.

Five Questions

1. The average prevalence of stuttering in the general population is:
   - **a.** 3 to 5 percent
   - **b.** 2 percent
   - **c.** one percent or less
   - **d.** 5 to 8 percent

   Answer: C

2. Data reported by Proctor et.al. (2008) showed that:
   - **a.** there was a greater incidence of stuttering among European American children than African American children.
   - **b.** there was a greater prevalence of stuttering among African American children than European American children.
   - **c.** there was a greater prevalence of stuttering among African American girls than African American boys
   - **d.** there was no difference in the prevalence of stuttering between African American children and European American children.

   Answer: D

3. The onset of stuttering occurs most frequently in which of the following age ranges:
   - **a.** 18 to 24 months
   - **b.** 25 to 48 months
   - **c.** 48 to 60 months
   - **d.** above 65 months of age

   Answer: B

4. Recent data pertaining to stuttering sub-types support:
   - **a.** Personality-based classification
   - **b.** Exterior/Interior stuttering-based
   - **c.** Developmental –based classification
   - **d.** Gender-based classification

   Answer: C

5. The percent of young children who stutter who have a family history of stuttering among either their immediate or extended family has been reported to be as high as:
   - **a.** 71%
   - **b.** 38%
   - **c.** 22%
   - **d.** 15%

Answer: A

Highlights

- Onset of stuttering
- Incidence and prevalence
- Developmental paths
- Genetic of stuttering
- Subtypes of stuttering

Table 1

Mean or Median (*) Age in Months at Stuttering Onset for 21st Century Studies

Study	Sample Size	M/F Ratio	Upper Age	Age at Onset		
				Total	Boys	Girls
Mansson (2000)	53	1.65	by age 8[1]	33	34.0	31.0
Buck, et al. (2002)	61	2.2	by age 7	36.3	35.1	39.1
Yairi & Ambrose (2005)	163	2.1	by age 6	33	33.6	32.9
Mansson (2005)	179	1.34	by age 4	30	30.5	29.6
Reilly, et al. (2009)	137	1.58	by age 3[2]*	29.9	29.3	29.7[2]
Howell, et al. (2008)	76	5.33	by age 12[3]	54.8	54.0	58.5

[1] All but two children began stuttering by 3 years of age.

[2] The gender breakdown was provided by Reilly in a personal communication (2012)

[3] The minimal age was 8.

Table 2

Stuttering incidence reported in 21st Century studies

Study	Sample size	Participants	Percent incidence
Mansson (2000)	1040	All children born during 2 years	5.09%
Felsenfeld et al. (2001)	3768	Adult Twins	8.80%
Craig et al. (2002)	12,131	Entire age range	3.22%
Månsson (2005)	928	92% of children born during 2 years	17.70 %
Dworzynski, et al. (2007)	12,892	Twin pairs ages 2–7	8.40
Reilly et al. (2009)	1,619	Preschool	8.50%

Table 3

21st century prevalence studies

Study	N	age	Prevalence	M/F Ratio
Okalidou & Kampanaros (2001)	1,113	4–5	2.20%	0.66
McLeod & Harrison (2009)	4,983	4.5	5.60%	
Proctor et al. (2008)	3,165	2–5	2.60%	2.6
McKinnon et al. (2007)	10,000	5–13	0.33%	
Van Borsel, et al. (2006)	21,027	6–20	0.58%	4.6
Craig, et al. (2002)	12, 131	2–99	0.72%	2.3
Boyle, et al. (2011)	119,367	3–17	1.60%	2.47

Table 4

21st Century Recovery Data

Study	Country	Recovery Rate
Mansson (2000)	Denmark	85%
Ryan (2001)	USA	68%
Johannsen (2001)	Germany	77%
Craig et al. (2002)	Australia	70%
Yairi & Ambrose (2005)	USA	76%
Mansson (2005)	Denmark	94%
Dworzynski, et al. (2007)	United Kingdom	87%
Howell, et al. (2008)	United Kingdom	50%
Howell & Davis (2011)	United Kingdom	52%