



Letter to the Editor

Stuttering prevalence, incidence and recovery rates depend on how we define it: Comment on Yairi & Ambrose' article Epidemiology of stuttering: 21st century advances
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In the article, “Epidemiology of stuttering: 21st century advances”, Yairi and Ambrose (Y&A) have reviewed research investigating incidence, prevalence and recovery from stuttering, and have concluded that the more recent research suggests that the lifetime incidence of stuttering may be substantially higher than the 5% figure that has frequently been cited in textbooks, and that the prevalence of stuttering in the population at large may be lower than the 1% figure frequently cited. This implies that a greater percentage of individuals may experience stuttering at some time in their lives than previously thought, and that recovery rates are greater than previously thought.

Y&A attribute the discrepancies in incidence estimates to a tendency of earlier (20th Century) studies to under-diagnose stuttering in early childhood, due to: (a) the reliance on retrospective reports from interviewees; and (b) an excessively restricted age range for monitoring of children in prospective studies. They then go on to argue that prospective studies, and in particular those that track children from 12 months upward, provide the most valid estimates. In forwarding this argument, they imply that the more vigilantly children are monitored for perceptible symptoms of stuttering, the more valid the resultant incidence estimates will be.

Insofar as we are content to define stuttering on a purely perceptual basis, and we agree on which perceptible symptoms should be considered as constituting early childhood stuttering, Y&A's argument appears reasonable. Unfortunately, not all of us do agree, and I would argue that the implications of the disagreement are more profound than Y&A's review suggests. In this letter I highlight three issues that I believe should be central to any consideration of incidence and prevalence, and which, I would argue, have not been adequately considered in the Y&A review. These are: (a) the minimum length of time symptoms need to persist in order to be classified as stuttering; (b) the subjective experiences of people who stutter (PWS); and (c) the need to distinguish between ‘stalling’ and ‘escape’ behaviors.

1. The minimum length of time symptoms need to persist in order to be classified as stuttering

On the basis of personal experience, Y&A (<http://dx.doi.org/10.1016/j.jfludis.2012.11.002>) suggest that, in retrospective studies, parents “simply would not think of reporting a 2-month severe stuttering episode that occurred years in the past”. In contrast, in prospective studies, such as Reilly et al. (2009), in which children are tracked from an early age and where parents are instructed to look out for symptoms of stuttering, Y&A suggest that it is far more likely that such brief episodes will be reported by parents, and therefore included in the calculation of stuttering incidence. Y&A imply, in their discussion of this issue, that a diagnosis of stuttering is already appropriate in cases where symptoms have persisted for two months (provided they are sufficiently severe). The position Y&A adopt in their review is fully in line with their earlier statement (Yairi & Ambrose, 2005, p. 27) that they “strongly disagree with the frequently expressed notion that a child must stutter for at least 6 months to be considered to stutter” on the basis that this would result in misleading scientific and clinical information about the nature of the disorder.

Although Y&A's objection to a 6 month minimum duration may be entirely reasonable, it does beg the question: Where should we draw the line? What about children who produce symptoms that last for only a week, or only a day – that then resolve, never to return? Should we include such children in our estimates of stuttering incidence? As Y&A make clear, the shorter the duration of symptoms required by a study for a positive diagnosis of stuttering, the higher the incidence of stuttering that it will compute. However, taken to the extreme, if the methodology adopted by a longitudinal study were

ever sufficiently thorough to enable it to include, in its incidence calculations, all brief episodes of speech containing a high proportion of stuttering-like disfluencies that occur during the life of the study, that study might conclude that the incidence of incipient stuttering in young children is close to 100%!

This question of minimum duration of symptoms required for a positive diagnosis is especially pertinent in cases where the symptoms are short-lived but relatively severe, and the importance of this issue is highlighted by findings that instances of sudden and relatively severe onset of stuttering symptoms are not uncommon (Yairi, 1983; Yairi & Ambrose, 1992, 2005). Unfortunately, none of the 21st Century studies cited in the Y&A review have clearly specified in their Methods sections a minimum duration for which symptoms needed to persist for a positive diagnosis, and it seems that, in most cases, a minimum duration criterion either did not exist or was never systematically applied. For example, in the Reilly et al. (2009) study (which reported a stuttering incidence of 8.5%), parents were instructed to telephone the researchers if they noticed stuttering symptoms appearing, and researchers then made a home visit to make a definitive diagnosis. This procedure may have resulted in the minimum duration of symptoms leading to a positive diagnosis being determined by the length of time that happened to elapse between the parents alerting the researchers to the appearance of symptoms and the researchers' carrying out a home visit and assessment. Reilly et al. (2009, p. 272) clarified that "Where possible, home visits were conducted within 2 weeks of the parent report" – suggesting that for some of the children in that study who received a positive diagnosis of stuttering, the duration of symptoms may have been under 2 weeks. In the recent Bornholm study (Månsson, 2007), all parents were interviewed and asked "Did your child stutter previously? Even short periods of stuttering are of importance" (p. 99). For a positive diagnosis, verification by two speech therapists was required at some point. However, no minimum duration criterion was cited, and although Månsson (2007) did state (p. 100) that "On average, the course of stuttering lasted 17.1 months", it is unclear in the write-up, how the start and end dates of the stuttering episode(s) were calculated. So again, it appears possible that for many of these children, the duration of their actual stuttering episode(s) may have been extremely brief.

Of course, ultimately there is no correct answer to the question of what the minimum duration of symptoms needed for a positive diagnosis should be, and whether or not we diagnose a child whose stuttering symptoms only persist for a day as a child who stutters (CWS) is entirely dependent on the definition of incipient stuttering we choose to adopt. However, when deciding on a definition of incipient stuttering, it would seem sensible to try to select the one that is likely to prove most useful. Arguably, with respect to incipient stuttering, the most useful definitions are those that best enable us to identify the children who are at increased risk of persisting in their stuttering and who are most likely to benefit from therapy. In this regard, as a general guideline, studies that show a large difference between childhood incidence and adult prevalence are likely to be less useful than studies where the difference is smaller. Thus for example, the definition used in Månsson's (2007) Bornholm study (that resulted in an incidence figure of 17.7%, recovery rate of 94.4%, and prevalence at the end of the study of about 1.1%), is likely to prove less useful than definitions (and associated diagnostic criteria) that lead to lower incidence and recovery rates being reported.

2. The subjective experiences of people who stutter

In the absence of a consensus regarding the exact nature of the mechanism(s) underlying stuttering, we have little choice but to base our diagnoses of the disorder on the presence of behaviors people typically perceive to be stuttering and/or on what Y&A refer to as "quantified speech characteristics" that correspond to those behaviors. With respect to young children, such perceptually-based diagnoses by necessity exclude the experiences of the children themselves (because they are not old enough to tell us what they are experiencing); whereas, in older children and adults, diagnoses may be strongly influenced by the speakers' self-reports of their own experiences. Indeed, in many borderline instances, the bottom line in determining whether or not adults are identified as people who stutter is whether or not they identify themselves as such. By and large, this boils down to whether or not they experience instances of 'loss of control' (Perkins, 1990) whereby they find themselves unable to initiate or complete motor execution of words in a timely manner, despite having no difficulty in accessing the form of those words in inner speech (see also the ICD9 definition; World Health Organization, 1977).

Although this topic has yet to be adequately researched, it seems likely that most adults who stutter are able to distinguish between disfluencies that result from execution difficulties (i.e. where they know exactly the words they wish to say) and disfluencies that stem from word-finding or formulation difficulties. They would not classify disfluencies stemming solely from word-finding or formulation difficulties as stuttering, even though, to an independent listener, such disfluencies might sound like stuttering. Similarly, most clinicians and researchers would probably consider word-finding difficulties, utterance-formulation difficulties, and stuttering to be three distinct and separate conditions, even though, in adults, all three may result in an increased production of stuttering-like disfluencies. In contrast, with respect to young children, there is no such consensus as to the nature of the difficulty they are experiencing when they appear to be stuttering. Indeed, researchers (e.g. Bernstein Ratner, 1997; Conture, Zackheim, Anderson, & Pellowski, 2004), have noted that (at least some of) the symptoms of incipient stuttering may stem from word finding or sentence formulation difficulties.

3. The need to distinguish between stalling and escape behaviors

With respect to the above three potential sources of stuttering-like disfluencies, it is noteworthy that speakers are only likely to engage in escape behaviors, such as rushing to complete words or using force to push words out, if they already

know exactly what words they want to say. In contrast, if speakers do not yet know what they want to say (because they are experiencing difficulty finding a word or formulating the utterance), they are not likely to resort to such escape behaviors. Rather, they are more likely to engage in stalling behaviors, such as repeating or prolonging whatever words or sounds they *can* say. Stalling behaviors help the speaker to avoid silent pauses (Howell & Sackin, 2001) and signal to the listener that the speaker is still trying to say something. They may thus also help the speaker to hold the floor. Many disfluency types (both stuttering-like and normal), including repetitions and prolongations can be produced both as escape behaviors and as stalling behaviors, and it may be difficult, indeed sometimes impossible to distinguish between the two. However, all other things being equal, escape behaviors are more likely to involve the use of force and a tendency to rush forward, whereas stalling behaviors are more likely to be relaxed (i.e. executed without excessive force or muscle tension) and unhurried (cf. Howell & Au-Yeung, 2002). Because of this, escape behaviors are more likely to result in rapid part-word repetitions, whereas stalling behaviors are more likely to result in (slower) repetitions and prolongations of whole-words, especially of words that are easy to produce. This notion, that single-syllable word repetitions more often constitute stalling than escape behaviors, is supported by findings from an fMRI study by Jiang, Lu, Peng, Zhu, and Howell (2012) which demonstrated that single syllable word repetitions are more likely to be accompanied by neural activity characteristic of normal disfluencies (typically produced at times of word finding difficulty) than that characteristic of other forms of stuttering-like disfluencies. It is noteworthy that the Stuttering Severity Instrument for Children and Adults (SSI-3; Riley, 1994) adopts diagnostic criteria that are fully consistent with the theoretical distinction outlined above, insofar as it states: “Repetition of one-syllable words may be stuttering if the word sounds abnormal (shortened, prolonged, staccato, tense, etc.); however, when these single-syllable words are repeated but are otherwise spoken normally, they do not qualify as stuttering” (p. 4).

Although, clearly, experimental verification is needed, the above reasoning suggests that, in young children, the sudden appearance of large numbers of easy, relaxed repetitions and prolongations is more likely to reflect the onset of a period of increased stalling due to word-finding or sentence formulation difficulty; whereas, the sudden appearance of large numbers of rapid part-word disfluencies accompanied by increased muscle tension and a tendency to rush forward is more likely to reflect the onset of a period of use of escape behaviors in response to difficulty with execution of words that have already been formulated.

This line of argument brings up an important, yet largely ignored, question. . . “What sorts of underlying impairment or difficulty do we want to include in our definition of incipient stuttering?” The widest possible definition would probably include any impairment or difficulty that results in raised levels of stuttering-like disfluencies, including motor-execution, word-finding, and sentence-formulation difficulties. A narrower definition might only include impaired ability to execute already formulated words. For example, the early, ICD9 definition of adult stuttering is of this type, insofar as it defines stuttering as “Disorders in the rhythm of speech in which the individual knows precisely what he wishes to say but at the time is unable to say because of an involuntary repetition, prolongation, or cessation of a sound” (World Health Organization, 1977, p. 202).

Y&A point out that the majority of researchers currently include single-syllable word repetitions in their list of symptoms indicative of stuttering. Be that as it may, this practice may nevertheless not be optimal. This is because, by increasing the likelihood of including disfluencies stemming purely from word-finding and sentence-formulation difficulties, the inclusion of single-syllable word repetitions may be obscuring important predictors of persistent stuttering (at least insofar as stuttering is defined in terms of the experience of loss of control and difficulty initiating articulation).

Y&A note the high concordance between what parents, teachers, SLPs and researchers interpret as stuttering, and deduce from this that the definition of stuttering is not an issue. Surely, however, this is a circular argument: parents will be guided by what SLPs and researchers tell them and, in the absence of an impairment-based definition, SLPs and researchers will be influenced by parents’ perceptions. The elephant in the room here is the fact that we do not yet know for sure the mechanism that gives rise to stuttered disfluencies. This continued uncertainty leads us to an unsatisfactory overreliance on perceptual factors as a basis for definition (See Jackson, Quesal, & Yaruss, 2012, for a discussion). We also need to remember that the colloquial use of the term “stuttering” does not in any way imply the existence of an underlying disorder or pathology.

Because of the ongoing uncertainty regarding the mechanism behind (the disorder of) stuttering, disagreements about how stuttering should be defined will probably continue for the foreseeable future. In light of this, it would perhaps make sense if future studies investigating incidence and prevalence were to make a point of explicitly stating, in their write-ups, the minimum length of time symptoms needed to persist in order to qualify for a positive diagnosis of incipient stuttering. It would also make sense for studies to provide two separate measures of incidence: one including, and one excluding, cases where stalling behaviors (and in particular relaxed whole-word repetitions) are the only symptoms. In this way there is an increased likelihood that the data they provide will remain valid and useful, if and when such definitional issues are finally resolved.

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