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Phonological disorders and stuttering in children: what is the frequency of co-occurrence?

MARILYN A. NIPPOLD

University of Oregon, Eugene, Oregon, USA

Abstract

In the profession of speech-language pathology, there is a strong belief that phonological disorders frequently occur in children who stutter. The purpose of this article is to examine recently published studies that addressed the frequency with which the two disorders co-occur. Over 10 years ago, a similar article was published in which studies that had been conducted from the 1920s through the 1980s were examined. Unfortunately, methodological problems with the earlier studies limited the conclusions that could be drawn. Because of the uncertainty generated by those studies, researchers since 1990 have continued to investigate this topic. Based on the previous review, it is argued that more rigorous methods are needed, including the use of more objective and comprehensive measures of phonological development and matched control groups of non-stuttering children. The current review indicates that frequency rates vary widely from one study to another, making it difficult to state with confidence just how often the two disorders co-occur. Possible explanations for the discrepancies are discussed, including differences in how phonological disorders are defined and identified. Suggestions are offered for future research to obtain more precise estimates of co-occurrence.

Keywords: Children, stuttering, phonological disorders, intervention.

Introduction

Some children who stutter also have a phonological disorder (Throneburg *et al.*, 1994; Paden and Yairi, 1996), just as some children who do not stutter have greater difficulty than their same-aged peers in mastering the sound system of their native language (Wolk *et al.*, 1993; Paden and Yairi, 1996; Shriberg, 1997). In the profession of speech-language pathology, it is widely believed that children who stutter are far more likely to have a phonological disorder than their non-stuttering peers (St Louis and Hinzman, 1988; Louko *et al.*, 1990; Wolk *et al.*, 1993; Bloodstein, 1995; Louko, 1995; Yaruss and Conture, 1996; Tetnowski, 1998; Yaruss *et al.*, 1998; Louko *et al.*, 1999). For example, it is often stated that 30–40% of children who stutter have a

Address correspondence to: Marilyn A. Nippold, Communication Disorders and Sciences, University of Oregon, Eugene, OR 97403, USA. e-mail: nippold@oregon.uoregon.edu

phonological disorder, in contrast to the 2-6% of children in the general population (Conture *et al.*, 1993; Bernstein Ratner, 1995; Louko, 1995; Wolk, 1998; Melnick and Conture, 2000).

The purpose of the article is to examine recently published studies that addressed the frequency with which the two disorders co-occur. Over 10 years ago, a similar article was published (Nippold, 1990) in which studies that had been conducted from the 1920s through the 1980s were examined. In that review, six out of eight studies reported a higher frequency of speech sound errors in children who stuttered compared to children who did not stutter (McDowell, 1928; Berry, 1938; Darley, 1955; Schindler, 1955; Morley, 1957; Andrews and Harris, 1964; Williams and Silverman, 1968; Seider *et al.*, 1982), and a survey indicated that 16% of children who stuttered also had a phonological disorder (Blood and Seider, 1981).

Unfortunately, methodological problems with the earlier studies limited the conclusions that could be drawn. These included the absence of matched control groups of children who did not stutter (Blood and Seider, 1981); the use of subjective testing and scoring procedures to examine speech sound development (McDowell, 1928; Schindler, 1955; Williams and Silverman, 1968); and the use of parental interviews, questionnaires, or informal observations to document phonological disorders rather than direct testing of children's speech (Berry, 1938; Darley, 1955; Andrews and Harris, 1964; Blood and Seider, 1981; Seider *et al.*, 1982).

Based on that review (Nippold, 1990), it is argued that more rigorous methods are required to investigate the frequency with which phonological disorders occur in children who stutter. Studies should include the direct testing of children's speech, using more objective and comprehensive measures that elicit the production of sounds in a variety of phonetic contexts at the isolated word level and in spontaneous conversational speech. In addition to assessing phonological development at the word level, assessment of spontaneous conversational speech is recommended in order to evaluate sound productions in a more naturalistic context, as performance on tasks requiring single word responses may not necessarily represent children's productions in connected speech (Hodson and Paden, 1991; Morrison and Shriberg, 1992; Smit, 1994; Bankson and Bernthal, 1998; Schwartz, 1998). Studies should also include control groups of non-stuttering children, matched to the stuttering children on age, gender, native language, dialect, cultural and ethnic background and socio-economic status. By definition, a phonological disorder can be said to exist only when such testing indicates that a child's speech sound patterns deviate from those of his or her peers living in the same speech community (Taylor, 1994; Schwartz, 1998). This is particularly relevant in pluralistic countries such as the USA, characterized by tremendous linguistic, cultural, and ethnic diversity.

Because of the uncertainty generated by the earlier studies, researchers since 1990 have continued to examine the frequency with which phonological disorders occur in children who stutter. In a science-driven profession such as speech-language pathology, the knowledge base must be as accurate as possible, particularly when beliefs about stuttering influence the direction of future research. For example, if the two disorders do have a high rate of co-occurrence, this could justify heightened efforts to investigate the underlying nature of the two disorders, including the possibility of a common origin. Interestingly, genetic factors have been linked both to stuttering (Felsenfeld, 1997) and to phonological disorders (Shriberg, 1997). Given the importance of this topic, this article reviews and analyses recently published studies that addressed the following question: How frequently do phonological disorders occur in children who stutter?

Literature review

In a pertinent investigation, Louko *et al.* (1990) questioned whether children who stutter are more likely to display phonological processes (e.g., gliding, stopping, cluster reduction) compared to their non-stuttering peers. They tested 30 children who stuttered and 30 children who did not stutter for the use of phonological processes in spontaneous conversational speech. Both groups had a mean age of 4:6 (years:months) (range = 2:5–6:11 for the stuttering group; range = 2:4–7:1 for the non-stuttering group), and contained 28 boys and two girls. The diagnosis of stuttering was based on a conversational speech sample, parent interview and administration of the Stuttering Severity Instrument (Riley, 1980). To examine phonological development, each child was audio- and video-taped for 30 minutes while talking and playing with his or her mother in a clinical setting, supplied with pictures, toys and objects. A sample of 300 words produced by the child was extracted from a 10 minute segment near the middle of the session, and analysed for phonological processes. Processes were identified using criteria from Edwards and Shriberg (1983), Grunwell (1982) and Stoel-Gammon and Dunn (1985).

The mean number of different processes produced per child was 3.4 (SD = 2.9) for the stuttering group and 2.3 (SD = 2.1) for the non-stuttering group. Although the raw numbers suggested an advantage for the non-stuttering group, an independent *t*-test indicated that the difference was not statistically significant (p > 0.05). A list of all processes used by the children in both groups was generated, and a total of 18 different processes were identified. The number of children in each group who exhibited each process was tabulated. A series of independent *t*-tests (with Bonferroni corrections) yielded only one statistically significant difference between groups, with cluster reduction being more common among children who stuttered. There were no statistically significant differences between groups for any of the other 17 processes (p > 0.01).

To further examine their data, Louko *et al.* (1990) classified each child as having 'disordered phonology' or 'normal phonology'. A child with disordered phonology used at least one 'atypical' or 'age-inappropriate' process during the conversational sample. An atypical process was defined as one that was seldom used by normally-developing children (e.g., glottal replacement) (Edwards and Shriberg, 1983); an age-inappropriate process was defined as one that persisted in speech beyond the typical age of resolution (Grunwell, 1982; Stoel-Gammon and Dunn, 1985). Using those criteria, they reported that 12 out of 30 (40%) children in the stuttering group had a phonological disorder compared with two out of 30 (7%) in the non-stuttering group, a result that was statistically significant (p=0.0006) according to a chi-square test.

This result is often cited in the literature to support the view that children who stutter frequently demonstrate phonological disorders (e.g., Conture *et al.*, 1993; Wolk *et al.*, 1993; Bernstein Ratner, 1995; Paden and Yairi, 1996; Yaruss and Conture, 1996; Louko *et al.*, 1999; Melnick and Conture, 2000). However, there are reasons to interpret it cautiously.

First, it is surprising that so many children in the stuttering group were identified as having a phonological disorder when the initial statistical analyses in this investigation indicated that the groups did not differ in the mean number of phonological processes per child, and that out of 18 different processes that were produced by the groups, only one (cluster reduction) occurred more often in the stuttering group. This result seems inconsistent with the subsequent report that six times as many stuttering children as non-stuttering children had phonological disorders. Second, the investigators themselves emphasised the preliminary nature of their study, mentioning the limitation that spontaneous conversational speech samples were used as the sole basis for identifying phonological processes. Because the children were free to talk about anything they wished, this procedure could not ensure that all children in both groups attempted to produce all English consonants and consonant clusters. The investigators recommended that future research involve a more thorough and systematic examination of phonological development. For example, sets of objects or pictures representing all sounds in the language, occurring in a variety of phonetic contexts, could be presented in a naming task designed to elicit those sounds. This procedure, combined with a lengthier conversational sample, would offer greater objectivity in identifying phonological disorders in children who stutter compared to their non-stuttering peers. Because of these limitations, it is reasonable to question the report that phonological disorders occurred with a much higher frequency in children who stuttered.

Yaruss *et al.* (1998) examined the diagnostic data of 99 children, ages 2 to 6 years (mean age = 4:7), who had been evaluated at a university speech and language clinic over a 12-year period (1978–1990). Each child had been referred to the clinic because of parental concerns that the child might be stuttering. A lengthy battery of speech and language tests had been administered to each child to determine the presence of stuttering and any other speech or language disorders. A child's use of phonological processes was examined during a picture-naming task and a spontaneous conversation between the child and parent. Using guidelines similar to those employed by Louko *et al.* (1990), described above, a child was said to have a phonological disorder if two or more atypical or age-inappropriate processes were exhibited. The investigators reported that 37% of the children had a phonological disorder.

Before accepting the conclusion that over one-third of the children who stuttered also had a phonological disorder, several issues should be considered. First, a matched control group of non-stuttering children did not participate in the study. This introduces subjectivity to the process of identifying phonological disorders, and makes it difficult to determine if the children were exhibiting actual disorders or normal variability in phonological development. Second, although Yaruss et al. (1998) appropriately employed two different contexts (picture-naming and spontaneous conversation) to identify phonological processes in the children's speech, few details were provided concerning the number of words and utterances that were elicited from each child, making it difficult to determine how thoroughly a child's phonological skills were examined. Third, there is reason to question whether all 99 children in the study were actually stuttering, particularly since treatment for stuttering was recommended for only 49 of them. The children were described as being 'at risk for stuttering' (p. 63), but a definite diagnosis of stuttering was not reported in this study. Perhaps some were showing normal disfluency, a behaviour that can resemble stuttering in children (Ramig and Shames, 1998). Without a clear diagnosis of stuttering, the statement that 37% of the children had a phonological disorder is impossible to interpret.

In another study designed to measure the frequency of phonological disorders in children who stutter, St Louis *et al.* (1991) examined the speech of 24 children whose stuttering had been identified during the National Speech and Hearing Survey (NSHS) (Hull *et al.*, 1976). From a database of nearly 39,000 children, the investigators randomly selected two children who stuttered from each of Grades 1 through 12 (ages 6.8-17.5 years; mean = 12.6 years). The resulting group consisted of 19 boys and five girls.

The investigators confirmed the diagnosis of stuttering by analysing the disfluencies produced by each child on an audio-recorded conversational speech sample. They also reviewed the audio-recordings of each child's picture-naming responses on the Goldman-Fristoe Test of Articulation (GFTA) (Goldman and Fristoe, 1968), which had been administered during the NSHS. Speech sound errors of type (omission, substitution, and distortion) and place (word initial, medial, and final) in isolated words were noted. As a group, the 24 children reportedly produced a mean of 3.4 errors. Each child was further classified as having either normal phonology (0–1 errors), or a phonological disorder that was mild (2–4 errors), moderate (5–9 errors), or severe (10 or more errors). Using those criteria, 58% of the children were reported to have normal phonology, and 42% were reported to have a phonological disorder. Of those reported to have a disorder, most were in the mild category (mild = 21%; moderate = 13%; severe = 8%). These results led the investigators to conclude that phonological disorders occur in a 'substantial proportion' (p. 13) of children who stutter.

Before accepting their conclusion, it is important to consider some limitations of this study. One is that no information was reported concerning the children's speech sound productions in spontaneous conversation. This information is necessary to verify the existence of phonological disorders in children (Morrison and Shriberg, 1992; Shriberg, 1997). Another problem is that the same classification system was applied to interpret every child's performance on the GFTA, regardless of chronological age or gender. Any child who produced two or more errors on the test was classified as having a phonological disorder, whether the child was 6 or 16 years of age. In a newer edition of the GFTA examiner's manual (Goldman and Fristoe, 1986), there are separate sets of norms for boys and girls of different ages, indicating that a child's performance on this test can be expected to vary in relation to age and gender. The criterion used to define a disorder as the production of two or more errors may have been too stringent for some of the children. For example, according to the manual (p. 25), a boy age 8:0 who produces two errors is performing at the 56th percentile, well within normal limits. It is also difficult to draw conclusions because of the small number of children participating in each grade level (n=2) and the absence of a control group that did not stutter. Because of these limitations, the report that 42% of the children in this investigation had a phonological disorder is subject to question.

Some of these limitations were addressed by Bernstein Ratner (1998) who also administered the picture-naming subtest of the GFTA (Goldman and Fristoe, 1986) to children who stuttered. The children in her investigation were much younger (mean age = 2:10) than those in the St Louis *et al.* (1991) study, and had been stuttering for only about 3 months. Each child's stuttering was documented in a conversational speech sample of at least 50 utterances. Each child who stuttered (n=12) was matched to a fluent control child (n=12) on the basis of age, gender, and socio-economic status. Results showed that both groups of children performed well within normal limits on the GFTA. In relation to the norms provided by the test, mean performance was at the 56th percentile for the children who stuttered, and at the 63rd percentile for those who were fluent. Differences between the groups were not statistically significant (p > 0.05). It should be noted, however, that a more thorough examination of the children's phonological development in the Bernstein Ratner (1998) study might have yielded a different outcome. A more comprehensive assessment that includes an examination of speech sound production in spontaneous conversation—in addition to isolated words—is likely to provide a more valid assessment of a child's phonological development (Hodson and Paden, 1991; Morrison and Shriberg, 1992; Smit, 1994; Bankson and Bernthal, 1998; Schwartz, 1998). Interestingly, Bernstein Ratner had elicited conversational speech samples from each child in the study but did not analyse them for phonological errors. It would be informative to revisit those samples, perform a detailed phonological analysis of each, and compare the performance of the groups.

Ryan (1992) conducted a similar investigation to Bernstein Ratner's (1998). He administered the Arizona Articulation Proficiency Scale (AAPS) (Barker, 1973) to 40 young children, half of whom stuttered and half of whom were fluent. The diagnosis of stuttering was confirmed through an analysis of disfluencies in the children's speech during a fluency interview. The two groups were matched for chronological age, gender, and number of years of education their mothers had completed. Children in the stuttering group had a mean age of 4:4 (range = 2:10–5:9), and those in the non-stuttering group had a mean age of 4:5 (range = 2:10–5:9). Mean scores on the AAPS were 91.4 (SD=7.5) and 94.2 (SD=6.8), respectively, for the stuttering and non-stuttering groups. Statistical analysis showed that the scores were not significantly different (p > 0.05). Moreover, both groups reportedly performed within the expected range in terms of the AAPS normative data. Hence, the study did not support the view that children who stutter are more likely to have phonological disorders than their non-stuttering peers.

It should be noted, however, that the AAPS is a screening tool, which examines consonant production in a limited number of phonetic contexts (word initial and final positions but not word medial positions). Moreover, as with the Bernstein Ratner (1998) study, the absence of data concerning the children's conversational speech leads to questions concerning the status of their phonological development. Had Ryan (1992) elicited a conversational sample from each child and analysed it for phonological errors, this would have permitted a more thorough examination of the children's sound productions and a comparison between stuttering and non-stuttering groups. It is possible that the procedure of relying entirely on a picture-naming task, particularly one that offers a cursory examination of speech sound productions, could overlook a child whose phonological difficulties are apparent only in conversation (Morrison and Shriberg, 1992).

Nevertheless, despite the non-significant group differences on the AAPS, Ryan (1992) reported that five children in the stuttering group, or 25% of the sample, eventually required therapy to correct residual speech sound errors when they were older. Residual errors are distortions of fricatives or liquids such as /s/, /r/, and /l/ that persist in children ages 9 years and older (Shriberg *et al.*, 1999). While this is an intriguing report, it is difficult to interpret because similar data for children in the non-stuttering group were not available. Research is necessary to determine the frequency with which residual speech sound errors occur in children who stutter compared to their non-stuttering peers. Ideally, longitudinal studies should be conducted, beginning during the toddler years and continuing through the schoolage years.

Discussion

In the introduction to this article, this question was posed: How frequently do phonological disorders occur in children who stutter? Consistent with the previous review (Nippold, 1990), the answer to this question is uncertain. Since 1990, five published studies have addressed this question, but collectively their findings are inconsistent. Three of the studies reported that a sizeable portion of children who stutter also had a phonological disorder, with figures of 40%, 42%, and 37% reported, respectively, by Louko et al. (1990), St Louis et al. (1991), and Yaruss et al. (1998). In contrast, Bernstein Ratner (1998) and Ryan (1992) reported that children who stutter did not differ from their non-stuttering peers in the frequency of phonological disorders. These inconsistencies call into question the widely held belief that children who stutter frequently have a co-occurring phonological disorder. It is clear that some children who stutter also experience phonological disorders, as do some children who do not stutter (e.g., Wolk et al., 1993). However, given the discrepancies with which the two disorders reportedly co-occur, the 30-40% figure that is commonly cited (e.g., Conture et al., 1993; Louko, 1995; Wolk, 1998; Louko et al., 1999; Melnick and Conture, 2000) may be inflated. In any case, even when children who stutter are reported to experience phonological disorders, those problems tend to be mild, affecting a small number of phonemes (St Louis et al., 1991) and laterdeveloping phonological patterns such as consonant clusters (Louko et al., 1990).

One source of variation across studies is the manner in which 'phonological disorder' is defined. For example, in one study (St Louis et al., 1991), a phonological disorder was defined as the presence of two or more errors on the GFTA (Goldman and Fristoe, 1968), regardless of the child's age or gender. In another study (Bernstein Ratner, 1998), that same test was employed but the normative data for age and gender contained in the technical manual were used to define a disorder. Another source of variation concerns the approach used to elicit speech to identify phonological disorders. Picture-naming tasks are often used to elicit the production of consonants in isolated words (St Louis et al., 1991; Ryan, 1992; Bernstein Ratner, 1998). However, some children may perform acceptably on such tasks, but display poor sound production in conversation (Morrison and Shriberg, 1992). In the absence of a comprehensive assessment of phonological development that includes a sample of spontaneous conversational speech-in addition to a comprehensive word production task—it is possible that children with subtle phonological disorders could be overlooked, resulting in the under-identification of this problem in those who stutter. Moreover, when studies fail to include a matched control group of fluent children (St Louis et al., 1991; Yaruss et al., 1998), it is difficult to determine if those who stutter are showing a disorder, delay, or simply normal variation in phonological development.

It should be noted that others have also questioned the association between stuttering and phonological disorders. For example, Throneburg *et al.* (1994) reported informally that from their own data pool of 75 young children who stuttered and 50 who were fluent, severe phonological disorders occurred in only 7% of those who stuttered and in 6% of those who were fluent, a diagnosis that was based on the children's performance on the Assessment of Phonological disorders were strongly related, a larger proportion of children who stuttered would be expected to have severe phonological disorders compared to their non-stuttering peers.

Because of the uncertainty surrounding this topic, additional studies are essential to obtain more precise estimates of the frequency with which the two disorders cooccur. In future research, the diagnosis of a phonological disorder should be based on a combination of measures that would permit a thorough examination of the child's speech sound productions in a variety of phonetic contexts in isolated words and in representative samples of spontaneous conversational speech. From an objective standpoint, a child can be said to have a phonological disorder only when performance on such tasks differs significantly from that of the child's true peers, taking into consideration key factors such as age, gender, native language, dialect, cultural and ethnic background, and socio-economic status (Taylor, 1994; Schwartz, 1998). This is particularly germane in pluralistic countries characterised by much linguistic, cultural, and ethnic diversity.

To enhance the validity of the results, large numbers of stuttering children should participate along with carefully matched control groups of non-stuttering children. The children's ages when they are examined should be considered as well. Although stuttering and non-stuttering groups may perform similarly during their pre-school years (e.g., Ryan, 1992; Bernstein Ratner, 1998), the possibility of residual speech sound errors requiring formal treatment during the school-age years must be considered (Rvan, 1992). Attention should be given also to the methods of recruiting children to participate in research to ensure that representative samples are obtained. There is no evidence to suggest that recruitment was a problem for any of the studies discussed in this article. However, it is reasonable to ask if children who stutter and have a phonological disorder are more likely to be referred to the speech-language pathologist than children whose only problem is stuttering, with the former child possibly thought to have 'a more serious speech problem', and hence a greater need for treatment. This itself is an important question for research: Why are some children who stutter referred to the speech-language pathologist while others are not? If research determines that those who have more than one speech problem are more likely to be referred and treated, this would bias the results of studies that examine the frequency of phonological disorders in children who stutter and recruit their participants from the speech-language pathologist's caseload. Other recruitment methods would be needed, such as screening entire pre-schools or day-care centres to identify children who stutter. Helpful guidelines for conducting large-scale studies of the prevalence of phonological disorders and their co-occurrence with other disorders can be found in Shriberg et al. (1999).

To conclude, phonological disorders in children who stutter has become a prominent topic in the literature. Although it is commonly believed that children who stutter frequently display a co-occurring phonological disorder, a careful analysis of recently published studies in this area reveals that the frequency with which the two disorders co-occur continues to be uncertain. Additional research is therefore essential to obtain more precise estimates of co-occurrence.

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