Stuttering in children: a literature review update

Gaguez na criança: atualização da revisão de literatura

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Abstract

Introduction: Stuttering is a fluency disorder in which the flow of speech is disrupted. The disorder is frequently misunderstood and to better analyze it is necessary to understand stuttering as more than a speech problem. Recent literature points out thatshould instead be viewed as a communication disorder with the potential to affect several aspects of children's lives. Different perspectives about stuttering can bring a more diverse analysis and move the field forward in scientific knowledge, however, it can also lead to fragmented and controversial views. Despite some lingering scientific consensus issues, there has been growing agreement among researchers that stuttering is a multifactorial disorder.
Aims: To summarize and analyze previously published research considering stuttering as a dynamic disorder influenced by several factors.
Materials and methods: A comprehensive review which focuses on the development of stuttering, and the implications for the onset, manifestation, and chronicity of this disorder in school-age children who stutter.
Results: Because of the ever-increasing literature in the area of stuttering, the review addresses assessment procedures and the perception of the impact of stuttering on children's daily life.
Conclusions: This comprehensive view contributes to an updated understanding of therapeutic and scientific factors to be considered in the evaluation and treatment of stuttering.

Palavras-chave
Gaguez; perturbações da fluência; crianças que gaguejam.

Resumo

Introdução: *A gaguez é uma perturbação da fluência na qual o fluxo da fala é interrompido. Para melhor compreender esta perturbação não é possível analisá-la como um simples problema de fala. Deve ser vista como uma perturbação da comunicação com potencial para afetar vários aspetos da vida das crianças. Diferentes perspetivas sobre a gaguez podem trazer uma análise mais diversificada e contribuir para o avanço científico nesta área; no entanto, também podem levar a visões fragmentadas e controversas. Apesar de nem sempre haver unanimidade científica entre os diferentes autores, existe um consenso crescente entre os investigadores de que a gaguez é uma perturbação multifatorial.*
Objetivo: *Resumir e analisar pesquisas publicadas anteriormente, considerando a gaguez como uma perturbação dinâmica, influenciada por vários fatores.*
Material e métodos: *Revisão da literatura focada no desenvolvimento da gaguez, manifestação e cronicidade em crianças com idade escolar que gaguejam. A revisão aborda ainda os procedimentos de avaliação e a perceção do impacto da gaguez na vida diária das crianças.*
Conclusões: *Esta visão abrangente contribui para uma melhor compreensão dos fatores terapêuticos e científicos que devem ser considerados na avaliação e intervenção da gaguez.*

Introduction

The human voice is a complex neurophysiological system supporting communication. Several mechanisms must interact in order for words to be produced efficiently and effectively. The passage of air from the lungs through the larynx leads to the vibration of the vocal folds. The resulting vibration is resonated and shaped by the lips, teeth, tongue, and other structures in the oral and nasal cavities.[1] To interact and talk with others, speakers put the sounds and words together, resulting in a forward flow of speech. Fluency is a term describing the continuity, smoothness, flow, and effort involved in the process of speech production.[2-4] Sometimes, for various reasons, this fluency can be disrupted. Stuttering is one of the resulting conditions that can be associated with disruptions in speech fluency.[5]

Development

The Stuttering Phenomenon

Stuttering can be described as a neurodevelopmental disorder that normally arises in young children.[6] It is characterized by interruptions of normal speech fluency. These interruptions may include repetitions of sounds, syllables and words; prolongations; blocks; and broken words. These disfluencies may be accompanied by secondary behaviors, such as involuntary movements of the limbs, head, lips, and eyes.[5,7] The disfluencies associated with stuttering are commonly called stuttering-like disfluencies.[8] People who stutter often exhibit negative feelings, thoughts and attitudes toward their speech and experience an adverse impact on the quality of life.[4,9]

Other types of disfluencies, such as hesitations, silent pauses, interjections of word fillers, nonword fillers, whole-word repetitions, and phrase repetitions, are also present in people who stutter, they are also common in people who do not stutter. This is especially true in young children. Thus, they are considered to be typical or non-stuttered disfluencies.[5,10]

Over the years, there have been attempts to define stuttering with a multidimensional view of the disorder; however, there are still some gaps in the definitions agreement. This reinforces the inherent complexity of stuttering. For example, research by Travis and colleagues, starting in the 1930s highlighted the neurophysiological basis of stuttering.[11] Also, Johnson's research was an important contribution to the field of stuttering. Probably, Diagnosogenic theory (1938) stating that: stuttering onset was related to the overreaction from parents to child's disfluencies, was the basis for the more in-depth studies on the influence of the environment.[12] However, the first theories of stuttering were based on a search for the cause, rather than a more dynamic approach, now advocated by many authors.[6,9,13,14] This is in line with Johnson's (1958) analogy of an elephant being examined through blind men, showing that stuttering can look and feel differently depending on the person who is seeing it.[15] Each blind man, who is examining the elephant, will come to different conclusions, as he is only examining a part of the animal.[16]

In 1970, Sheehan proposed an analogy for understanding the multidimensional nature of stuttering: "Stuttering is like an iceberg, with only a small part above the waterline and a much bigger part below".[17] Despite unidimensional treatment is still widely used, the idea of treating stuttering through simple fluency control began to be break up, and over the years, the concept of multidimensionality in stuttering has become increasingly accepted. Accordingly, several models have been developed to describe the ways in which different aspects of a person's experience might combine in the experience of stuttering.[18-25]

Perkins (1990) moved away from definitions related to the features of stuttering that might be observable to a listener and emphasized instead the speaker's underlying experience of stuttering. Perkins emphasized that stuttering should not be defined by observable behaviors but rather by the speaker's judgment of the loss of control in the ability to perform speech fluently.[26] Current research on stuttering has supported the loss of control as part of how speakers experience stuttering.[27]

Relatively recent, there have been efforts to understand stuttering in light of the International Classification of Functioning, Disability, and Health (ICF). This has led to new perspectives on stuttering, with a major focus on the impact of the disorder in people's lives. ICF describes all health-related experiences in terms of body structure and function, as well as activities and participation, including contextual factors.[28] According to the model, the analysis of stuttering components should include: a) the etiology; b) disability in body function (observable characteristics of stuttering); c) cognitive, behavioral, and affective reactions of the speaker towards stuttering; d) the influence

of the environment on stuttering (e.g., difficulties in speaking in different situations), and; d) the overall impact of stuttering on the person's life (indicated by limitations in communication activities and restrictions on participation).[4,27,28]

The DSM-V definition of stuttering takes into account some of the functionality concepts mentioned above.[29] Stuttering is defined as "childhood-onset fluency disorder." In addition to describing the surface features of stuttering, the DSM-V points out that the disorder may interfere with academic or professional success or social communication.

Although there are some controversies in the different definitions of stuttering, current definitions and theories increasingly encompass components that go beyond superficial features. This reinforces the need to ensure that therapeutic programs address the entire stuttering disorder, not just the surface characteristics.

In addition to the research that has been developed with a focus on the multidimensionality of stuttering, several studies have sought to explain the factors that underlie spontaneous recovery and chronicity of stuttering.[5,30,31] Typically, children who stutter begin to show symptoms between the ages of 2½ and 4 years old. Importantly, somewhere between 50% and 90% of those children recover from stuttering spontaneously or with treatment.[31,32] Stuttering tends to persist in children with specific risk factors that can also influence the beginning, maintenance, or severity of stuttering.[31,33] Research on those risk factors is ongoing.[13,34-36]

A Multifactorial Disorder

Currently, stuttering is considered a multifactorial disorder, which means that there is believed to be no single cause. Instead, several factors are believed to interact in unique ways to result in stuttering.

There is plenty of evidence about the existence of a genetic predisposition. Research has identified genetic mutations associated with stuttering;[37-40] however, there are, as yet, no definitive results regarding transmission models, chromosomes, genes, or sex factors that are involved in genetic expression.[39,41] This genetic predisposition can be triggered by neurophysiological factors, environmental, temperament, and language development.[7,42] According to Smith and Weber (2017), stuttering results from the instability of neural networks and their relationship to the environment. The breaks in the flow of speech may lead to responses in the child's internal and external environment, and this, in turn, can lead to behavioral and physiological changes. These processes may have epigenetic influences in the expression of genes involved in speech motor and other aspects of development.[6]

This view is consistent with the Dual Diathesis – Stressor model (DD-S) of developmental stuttering.[43,44] The DD-S model is a relatively recent framework that proposes that the endogenous abilities of children who stutter (*diatheses*) interact in a dynamically way with exogenous contexts *(stressors)*. The model is consistent with the view that stuttering involves emotional and cognitive components in addition to speech production differences.[6, 9,45-48]

Endogenous abilities play an important role in the development of stuttering,[43] and there is a growing scientific interest in how temperament[49] and executive functions[56] may influence this process. Therefore, these abilities can be thought of as a part of the *diathesis* that may contribute to the stuttering phenomenon.[43]

Temperament studies have concluded that children who stutter are more likely to be reactive and sensitive compared to their nonstuttering peers,[46] with a tendency for impulsivity.[48,56] Some studies have also reported that children who stutter have difficulty adapting to new objects and situations,[50] exhibit a greater negative effect,[51] and may have difficulties in self-regulation.[52] Temperament and EF have common strands; however, they have been investigated separately.[53] For example, certain temperamental characteristics, such as attentional focusing,[54] are assumed to have cognitive underpinnings in the executive or anterior attention system.[53,55]

Previous studies reported that children who stutter are less successful in maintaining attention[56-58] and selecting information from sensory input.[59] Findings also indicate a tendency for impulsivity compared to nonstuttering peers.[60–62] In addition, studies indicate that children who stutter perform less well in working memory than their nonstuttering peers.[63,64] Reflecting on the close relationship between cognition and language, it is interesting to note prior studies reporting differences in children's speech sound[65-68] and more advanced language skills.[69]

Environmental factors interact with intrinsic factors, such as temperament and cognitive abilities. Over time, this may lead to the development of unhelpful thoughts, negative emotions, and consequently, anxiety.[70,71,43] This is in line with several studies reporting that conditions with a genetic predisposition,

such as anxiety, and depression, become more likely in the presence of negative life stressors.[72,73]

Anxiety has been associated with stuttering, yet its relationship with stuttering is still controversial.[74-76] Although several reports indicate that adults who stutter experience elevated levels of anxiety,[74,77,78] people who stutter do not necessarily have to be anxious, however, they may experience anxiety in social situations involving speech.[45,75,76,79,80]

Currently, the occurrence of anxiety in children who stutter is still the subject of strong debate. Although some studies indicate a higher level of anxiety in children who stutter, other studies suggest that anxiety tends to manifest more clearly in older children, with a tendency to increase over time.[75,79–82] Some other studies have found no specific trend toward elevated anxiety in children.[83–86]

Anxiety can be influenced by how people who stutter see themselves. This may be related to their internal abilities, such as cognitive and temperament traits. As described in the next section, when combined with the attitudes and reactions of other people, anxiety may lead to restrictions in a person's daily activities and participation in society.[87]

Stuttering Assessment

Assuming that stuttering is a multifactorial disorder, with several aspects that may influence the onset, manifestation, and chronicity of the condition in each individual child, the assessment process should also include several components. This will allow clinicians to understand the whole disorder and see how it affects the daily life of the child.[6,9]

Over the years, numerous measures have been developed to assess the stuttering disorder. Some of these measures are primarily focused on the observable features of stuttering, while others address feelings, attitudes, thoughts, and reactions. For example, the *Stuttering Severity Instrument* (SSI4)[88] measures severity based on frequency, duration of stuttering moments, physical concomitants, and naturalness, for children and adults. One of the challenges facing such measures is the variability of stuttering in the results; however, it is still among the most used instrument in scientific research.[76,89,90.]

The *Communication Attitude Test* (CAT)[91] and the *Overall Assessment of the Speaker's Experience of Stuttering – Ages 7-12* (OASES-S)[9] are good examples of measures intended to assess more than the visible features of stuttering, such as how children react to stuttering.

The CAT is a self-report instrument that includes 35 true/false statements about speech-associated attitudes of school-age children who stutter. Psychometric measures show that CAT is a valid instrument with strong reliability which can be used in research and clinic to evaluate how children think and feel about their stuttering.[91-93]

The OASES-S is another self-report instrument. It is based on the WHO's ICF as adapted to stuttering by Yaruss and Quesal.[3,4] The sections of the OASES each relate to specific aspects of the ICF. There are three versions of the OASES: the OASES-A for adults, ages 18 and above; the OASES-T for teenagers, ages 13–17; and the OASES-S for school-age children, ages 7–12. The OASES instruments have shown good reliability and validity in the original English version,[9] as well as in different translated versions around the world. This shows that the OASES is a suitable instrument for both clinical and research use that can be used to collect information about the impact of stuttering in the lives of children, adolescents, and adults who stutter.[94-105]

Because children may not have a full understanding of the ways in which stuttering might affect them, it is also important for clinicians to gather information from parents and other relevant people. This can be done through informal interviews or through formal scales, such as the *Palin Parent Rating Scales,*[106] and observational rating scales, such as those included in the *Test of Childhood Stuttering* (TOCS).[107] Other forms designed to collect comprehensive history can be found in books and in treatment programs.[7,25,108-111]

Stuttering Impact

The person who stutters may experience negative affective, behavioral, and cognitive reactions from himself and from the environment. These can interfere in the individual's ability to participate in daily activities, including schoolwork. It may also affect their professional choices, interpersonal relationships, mental health (including the potential for increased social anxiety), and more.[9,76, 93,112,113]

Limitations from stuttering are not the same for everyone. This may be related to an individual's experiences of stuttering. Importantly, the degree of adverse impact a person experiences is not necessarily related to the observable severity of the disorder.[4,114]

The way that society perceives stuttering can also contribute to the impact of stuttering in people's lives. Although there have been some changes in

society, there are still strong negative stereotypes about stuttering. These negative attitudes can even be found in people responsible for education and employment opportunities.[115-118] Some stereotypes, beliefs, and attitudes are consistent across countries, while other beliefs are regionally or culturally specific. This is especially true for beliefs related to religious causes.[118] For example, the findings of Valente and colleagues with the Public Opinion Survey of Human Attributes-Stuttering (POSHA-S), revealed notable differences between countries and cultures across Europe.[119]

According to several reports, children are aware of their stuttering shortly after the onset. As they grow, the impact of the disorder may increase.[120,121]

The school-age and adolescent years are important for the development of cognitive processes and executive domains responsible for information-processing, cognitive flexibility, and goal-setting. During this time, children who stutter often have negative experiences at school.[71,122–124] Children at this age usually have already accumulated several years of experience with stuttering, and this can result in avoidance behaviors, as well as negative thoughts and emotions. These can influence and be influenced by interactions with others, especially those closest to children: parents and teachers.[125,126]

Despite common historical beliefs, it is currently known that emotional problems and parental style do not cause stuttering. Nevertheless, the ways in which people in the child's environment cope with and react to the disorder can influence children's emotional reactions, and avoidance behavior.[45,127] The coping patterns and styles of people in the child's environment, such as parents, and teachers, are influenced by the way they see the disorder and by the different ways the stuttering can affect children.

Understandably, parents may be worried about their child's speech. Such concerns may be related to the beginning of school, the possibility of bullying and other negative experiences at school, and to fears about the child's future[128,129,130] and revealed that parents are aware of the impact of stuttering on children's quality of life and of the difficulty their children may experience in communicating freely. However, to date, there have been few studies comparing the children's and parents' views regarding the impact of stuttering.[130,131]

Apart from the importance of analyzing the perspective of parents' impact, it is also fundamental to analyze the perspective of other individuals who spend time with the children in other settings, such as teachers. One reason that this is important is the variability of stuttering: people may stutter more or less in different situations.[45,132] School-age children divide most of their daily time between home, and at school with their teachers and classmates,[112,133] so the perspective of teachers is particular important.

Some reports highlight the negative perceptions and stereotypes held by teachers regarding people who stutter.[134,135] Other studies highlight a general lack of knowledge.[136–138] Such findings highlight the need to improve teamwork between professionals.

Conclusion

The misunderstandings that remain in the field of the stuttering result, in part, from different perspectives that have historically focused only on one part of the problem. Stuttering is a complex disorder including numerous factors that may be similar across individuals; however, individual differences also play an important role in the development of stuttering.

This review highlighted the importance of analyzing, and address into therapy, all aspects of the stuttering disorder, including not only fluency enhancement but also cognitive and social aspects. A multidimensional approach is essential for the evaluation and treatment of children who stutter. This assessment should include all major contexts in which children spend time, as well as all of the key people children, encounter in these contexts. This particularly means that parents and teachers should play a central role in the evaluation of stuttering to reduce the impact of stuttering on the child's life and minimize the negative impact that the child may already experience. To accomplish this, speech therapists should consider the cognitive, emotional, and social aspects of stuttering and establish good partnerships with parents, teachers, and other relevant people in treatment for children who stutter.

Bearing in mind the importance of the theme addressed, it would be relevant to carry out in the future a systematic review of the literature, in order to analyze in a deeper and more detailed way all the scientific evidence about this topic.

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