UNIVERSIDADE CATÓLICA PORTUGUESA

School age Portuguese children who stutter: socio - cognitive performance and the impact of stuttering on their quality of life

Tese apresentada à Universidade Católica Portuguesa
para obtenção do grau de Doutor em Ciências da Cognição e da Linguagem

Por

Mónica Filipa Soares Rocha

Instituto de Ciências da Saúde

Setembro 2019

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Sob orientação de:
Professora Doutora Joana R. Rato
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It's easier to build strong children than to repair broken men (Frederick Douglas)

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Resumo

A gaguez é uma perturbação da fluência que tem vindo a ser descrita, por alguns autores, como uma perturbação do neurodesenvolvimento. Resulta da interação entre diversos fatores, tais como genéticos, neurofisiológicos e ambientais e é caraterizada por interrupções ou bloqueios na fluência da fala, acompanhada, frequentemente, de esforço e/ou comportamentos de evitamento. A forma como os diversos fatores, associados à gaguez, interagem entre si ainda não é totalmente conhecida e continua a ser alvo de discussão. Compreender o desenvolvimento da gaguez ao longo de diferentes faixas etárias, bem como os fatores de risco que podem estar na base da recuperação espontânea ou cronicidade da perturbação é o que tem despoletado o interesse da investigação nesta área.

Nos últimos anos, vários autores mostraram que o impacto da gaguez não se limita às disfluências observadas na fala, existindo fortes evidências sobre os efeitos psicossociais da perturbação, não apenas na pessoa que gagueja, mas também junto de outros intervenientes, quer do seio familiar, quer do seio escolar.

O presente trabalho apresenta um conjunto de estudos, cujos principais objetivos visam compreender a interação entre alguns dos fatores, que são apontados na literatura como relevantes para o desenvolvimento da gaguez ou que se relacionam, de alguma forma, com essa perturbação. Pretendeu-se ainda identificar o impacto da gaguez, no quotidiano das crianças Portuguesas, comparando a sua autoperceção com a de crianças de outros países. A perspetiva de impacto dos pais e os dos professores das crianças foi igualmente estudada. Participaram 100 crianças Portuguesas, em idade escolar (7-12 anos; M=9.13; SD=1.70), das quais 50 crianças gaguejavam e a outra metade correspondia aos seus pares fluentes. A amostra correspondente às crianças que gaguejam foi recolhida por conveniência, em diferentes cidades de Portugal, através de casos referenciados por profissionais de saúde ou de educação. O diagnóstico de gaguez foi confirmado através da utilização do *Stuttering Severity Instrument* (SSI4). Nos estudos com outros intervenientes, participaram ainda 50 pais e 27 professores das crianças do grupo de crianças que gagueja.

São cinco os capítulos que constituem o presente trabalho de investigação. No primeiro, apresenta-se uma breve revisão da literatura que tem como base a

multidimensionalidade inerente à gaguez e onde se atualizam os conceitos, as causas e o impacto da perturbação na vida das crianças.

De seguida, apresenta-se o primeiro estudo, cujo objetivo consistiu em relacionar a gaguez com o temperamento, o funcionamento executivo e a ansiedade, através da comparação de crianças que gaguejam e os seus pares fluentes. O temperamento foi avaliado através do questionário parental: *Temperament in Middle Childhood Questionnaire* (TMCQ), traduzido e adaptado para este efeito. A avaliação do funcionamento executivo foi realizada através da versão portuguesa do instrumento neuropsicológico *Children Color Trails Test* (CCTT). A ansiedade foi avaliada através da versão portuguesa do questionário de autoperceção *Multidimensional Anxiety Scale for Children* (MASC). Neste estudo, os grupos de crianças que gaguejam e que não gaguejam foram divididos em dois subgrupos, tendo em consideração a idade: o grupo de crianças mais novas (7-9 anos); e o grupo de crianças mais velhas (10-12 anos). Os resultados indicaram que as crianças que gaguejam podem precisar de mais tempo na orientação da atenção e, tendencialmente, manifestam maior reatividade emocional, em comparação com seus pares fluentes.

Contudo, quando é considerada a idade das crianças, observam-se resultados significativos na comparação dos diferentes domínios de temperamento, apenas no subgrupo de crianças mais velhas, com médias superiores para as crianças que gaguejam, nas subescalas de: Raiva/Frustração; Impulsividade e Tristeza; e médias inferiores nas subescalas de: Atenção/Foco; Sensibilidade Percetiva e Recuperação de picos de Excitação. Por sua vez, no subgrupo de crianças mais novas, é no desempenho na tarefa relacionada com o funcionamento executivo que se verificam diferenças estatisticamente significativas. Neste grupo, as crianças que gaguejam utilizaram mais tempo a executar a tarefa, cometendo também um maior número de erros e falhas de execução, em comparação com os seus pares fluentes. Relativamente à ansiedade, e fazendo a mesma comparação entre os grupos de crianças que gaguejam e os seus pares fluentes, não foram encontradas diferenças significativas. Por último, foram encontradas correlações positivas entre a idade, alguns dos domínios relacionados com o funcionamento executivo e temperamento, sugerindo interação entre os diferentes fatores. Estes resultados permitiram reforçar a multidimensionalidade da gaguez e podem ser interpretados à luz do modelo *Dual Diathesis - Stressor* (DDS) que propõe que as capacidades endógenas (*diathesis*) das crianças interagem de forma dinâmica com os contextos exógenos (*stressor*).

O segundo estudo pretendeu medir o impacto da gaguez na vida das crianças Portuguesas e verificou uma perceção de impacto moderado da gaguez nas suas vidas. Foi utilizado o questionário de autoperceção de impacto *Overall Assessment of the Speaker's Experience – School age children* (OASES-S), que tem como base a Classificação Internacional de Funcionalidade (CIF) e que neste estudo foi traduzido, adaptado e validado para Português Europeu (OASES-S-PT). Verificou-se que o OASES-S-PT apresenta características psicométricas de fiabilidade e validade adequadas para avaliar a autoperceção de impacto nas crianças Portuguesas em idade escolar. Na comparação dos resultados com outros países, os resultados demonstraram que as crianças Portuguesas experienciam um impacto semelhante ao experienciado pelas crianças Holandesas e menor que as crianças Australianas. Verificou-se ainda um impacto menos relevante no quotidiano das crianças que gaguejam, comparando com as crianças Americanas, Holandesas e Australianas.

De forma a perceber a perspetiva dos pais e professores, relativamente ao impacto da gaguez na vida dos seus filhos e alunos, respetivamente, foram realizados os últimos estudos, com os objetivos de comparar a autoperceção das crianças com a perceção dos seus pais e professores. Para estes estudos foram realizadas adaptações das questões do instrumento OASES-S-PT de forma a poderem ser respondidos por pais (OASES-S-P) e professores (OASES-S-T).

No estudo direcionado para a perceção dos pais, realizou-se uma subdivisão do grupo, tendo em consideração a presença/ausência de história familiar de gaguez. Os resultados globais dos pais foram semelhantes aos resultados das crianças. No entanto, no caso dos pais com história familiar de gaguez, verificou-se maior impacto nos parâmetros relacionados com as reações das crianças à gaguez e a sua qualidade de vida.

No estudo com os professores, apesar de não se terem verificado diferenças significativas na comparação das respostas com as dos seus alunos, não se observaram correlações entre as respostas dos dois grupos. Para além disso, identificou-se uma elevada taxa de absentismo num conjunto de questões sobre a gaguez em geral e intervenção terapêutica. Estes resultados sugerem que, apesar dos professores terem consciência de que a gaguez pode afetar a vida das crianças que gaguejam, as suas respostas não estão em total consonância com as dos seus alunos e denunciam desconhecimento em determinados domínios. É discutida a importância da educação dos professores relativamente à gaguez, bem como da ação destes principais intervenientes que acompanham as crianças que gaguejam.

Os estudos realizados vêm reforçar a necessidade de desenvolver programas terapêuticos mais ajustados às particularidades de cada grupo específico, assim como destacar a importância do trabalho de parceria, quer com familiares, quer com professores. Contribui também para destacar a visão multidimensional, ressaltando a mais-valia na utilização de protocolos de avaliação que tenham em consideração outros domínios (ex: cognitivos, temperamentais, sociais), para além da fluência da fala.

Conclui-se que a dessensibilização das crianças que gaguejam para a perturbação pode ajudar a lidar de uma forma mais positiva e menos reativa à gaguez, facilitando o processo terapêutico. Assim, um processo de avaliação e intervenção mais efetivos podem ser decisivos para a diminuição do impacto que a gaguez possa causar na vida das crianças.

Abstract

Stuttering is a multifaceted disorder resulting from the interaction between genetic, neurophysiological, environmental, and other factors. The ways these factors interact with each other is not yet fully understood, and is still subject of research. In recent years, several authors have shown the impact of the disorder to reach far beyond the surface components, with strong evidence about the psychosocial effects of stuttering not only for the person who stutters but also for their family.

This research work seeks to clarify the interaction between factors that contribute to the development of stuttering. In particular, it attempts to improve the assessment and therapeutic programs for stuttering in Portugal. This work presents five chapters: the first consists of a literature review based on the multidimensionality inherent to stuttering. The following chapters consist of four studies that are detailed below.

The first study aimed to relate stuttering to temperament, executive functioning, and anxiety, by comparing stuttering children and their nonstuttering peers. Participants were 100 school-age children (mean age = 9.13 mos.; SD = 1.70 mos.), which included 50 children who stutter and 50 children who did not. In this study, participants were divided into two subgroups, taking age into account: a younger group (7-9 years) and an older group (10-12 years). Their temperament was evaluated through the parental questionnaire, *Temperament in Middle Childhood Questionnaire (TMCQ),* translated and adapted for this purpose. Executive functioning was assessed through the Portuguese version of the neuropsychological instrument, *Children Color Trails Test (CCTT).* Anxiety was assessed through the Portuguese version of the *Multidimensional Anxiety Scale for Children (MASC)* self-assessment questionnaire.

The second study aimed to measure the impact of stuttering on the Portuguese children's lives. Participants were 50 children who stutter (Mean age = 9.10 mos.; SD = 1.7 mos.). For this study, the *Overall Assessment of the Speaker's Experience of Stuttering - School age* (OASES-S) (OASES-S) was translated, adapted, and validated for European Portuguese (OASES-S-PT). Good reliability and validity of the OASES-S-PT were verified for the assessment of impact in Portuguese school-age children.

The third and fourth studies were conducted in order to understand the parents' and teachers' perspectives regarding the impact of stuttering on the lives of their children and students, and to compare the results with the children's self-perception of impact.

In the study related to the perception of parents, a subdivision of the group was performed, taking into account the presence or absence of a family history of stuttering. The participants were 50 parents divided into: i) group of parents with a family history of stuttering (n= 30), and ii) group of parents with no history family of stuttering (n= 20). The study that aimed to investigate the teacher's perceptions involved 27 teachers. In both studies adaptations, of the OASES-S-PT instrument were made so that they could be answered by parents (OASES-S-P) and teachers (OASES-S-T).

The results indicated that children who stutter may experience lower ability to orient attention and may have greater emotional reactivity compared with their nonstuttering peers. Portuguese children who stutter experience a moderate impact of stuttering in their lives. In general, the results of parents and teachers were in agreement with the perceptions of the children; however, parents with a family history of stuttering reported significantly higher negative impact. This finding supports the idea that refinements in clinical and educational practices are needed; findings also highlight the importance of teamwork with parents and teachers.

List of abbreviations and acronyms

CBQ - Children's Behavior Questionnaire

CCTT - Children's Color Trails Test

CAT - Communication Attitude Test

DD-S – Dual Diathesis – Stressor

EF – Executive Functioning

ICF - International Classification of Functioning, Disability, and Health

MASC - Multidimensional Anxiety Scale for Children

OASES - Overall Assessment of the Speaker's Experience of Stuttering

OASES-S - Overall Assessment of the Speaker's Experience of Stuttering – School age children

OASES-S-PT - Overall Assessment of the Speaker's Experience of Stuttering – School age children – European Portuguese Version

OASES-S-PT-P - Overall Assessment of the Speaker's Experience of Stuttering – School age children – European Portuguese Version – Parents' adaptation

OASES-S-PT-T - Overall Assessment of the Speaker's Experience of Stuttering – School age children – European Portuguese Version – Teachers' adaptation

PCA - Principal Component Analysis

POSHA-S - Public Opinion Survey of Human Attributes-Stuttering

SPSS - Statistical Package for the Social Sciences

SSI 4 - Stuttering Severity Instrument – 4th Edition

TMCQ - Temperament in Middle Childhood Questionnaire

TOCS -Test of Childhood Stuttering

WHO - World Health Organization

Table of Contents

INTRODUCTION

Stuttering is a communication disorder characterized by involuntary interruptions in the flow of speech, which encompasses repetitions; prolongations; blocks, and broken words (Bloodstein & Bernstein Ratner, 2008). Most of the children who begin to stutter will spontaneously recover from stuttering. This fact has led to numerous studies comparing children who experienced natural recovery and children who continue to stutter (Bloodstein & Bernstein Ratner, 2008; Chang et al., 2018; Franken, Koenraads, Holtmaat, & van der Schroeff, 2018; Smith & Weber, 2017; Yairi & Ambrose, 2005).

It is clear that stuttering has a genetic and neurological basis (Alm & Risberg, 2007; Chang et al., 2018; Kraft & Yairi, 2011; Kronfeld-Duenias, Civier, Amir, Ezrati-Vinacour, & Ben-Shachar, 2018; Neef et al., 2018; Smith & Weber, 2017). In addition, areas such as executive functioning, and temperament are increasingly being investigated, reinforcing the idea of dynamism in the origin and development of the disorder (Smith & Weber, 2017).

There has been an effort to investigate beyond the surface features of stuttering (Beilby, 2014; Erickson & Block, 2013; Manning & Beck, 2013; Vanryckeghem, Hylebos, Brutten, & Peleman, 2001; Yaruss & Quesal, 2016); however, there are still gaps in the literature in relating social, emotional, and cognitive domains.

Over time, much has been written about the increased risk for developing social and psychological problems, in children who stutter (Craig, Hancock, Tran & Craig, 2003; Craig, Blumgart, & Tran, 2011; McAllister, Kelman, & Millard, 2015; Neumann et al. 2017; Vanryckeghem et al, 2001).

The negative impact of the disorder can be seen in different domains, such as daily activities, relationships, academic performance, and mental health (e.g. anxiety) (Craig, Blumgart, & Tran, 2009; Koedoot, Bouwmans, Franken, & Stolk, 2011; Yaruss & Quesal, 2016). All these impacts on quality of life are represented in the World Health Organization [World Health Organization (WHO), 2001] framework, the International Classification of Functioning, Disability and Health's (ICF).

There is a growing consensus that the adverse impact of stuttering is not only related to observable speech. The present research is motivated by the idea that the intrinsic factors for each person (such as temperament, executive functioning), together

with the external factors (such as the children's environment) are crucial cornerstones that combine to increase or diminish the impact of stuttering in children's lives (Walden, Frankel, Buhr, Johnson, & Karrass, 2012). How other people, especially those who are closest to the child, such as parents and teachers, react to stuttering play an active role in this inherent dynamism of stuttering (Alm, 2014; Berquez & Kelman, 2018).

This dissertation has socio-cognitive performance and stuttering impact as its central theme and pursuit to answer the main research questions:

i) *Do Portuguese children who stutter show significant differences compared to their nonstuttering peers in measures of temperament?*

ii) *Do Portuguese children who stutter show significant differences in relation to their nonstuttering peers in measures of executive functioning?*

iii) *Do Portuguese children who stutter show significant differences in relation to their nonstuttering peers in measures of anxiety?*

iv) *Is there any correlation between temperament, executive functioning, and anxiety in children who stutter?*

v) *What is the impact of stuttering on quality of life of Portuguese Children who stutter?*

vi) *How do the parents' perceptions of the impact of stuttering on their children compare to the children's own perceptions?*

vii) *Does the teachers' perspective of the impact of stuttering on the life of his/her student is similar to the child's own perspective?*

To address these questions, the main objectives are:

i) To analyze temperament, executive functioning, and anxiety in children who stutter, and compare their performance with children who do not stutter; ii) correlate the results of temperament, executive functioning, and anxiety in the children who stutter; iii)

translate, and adapt the OASES: Overall Assessment of the Speaker's Experience of Stuttering - School age (OASES-S; Yaruss & Quesal, 2016b) (Annex 1) for European Portuguese iv) explore the impact of stuttering in children who stutter and compare it with impact assessed in other countries; v) investigate the impact of stuttering in Portuguese school-age children from the parents' and teachers' perspective, and compare it with the children's own perspective.

In order to attain all issues and aims, a literature review was performed, and four studies were implemented. Thus, the content of this dissertation is composed in a total of five chapters:

- Chapter 1: Stuttering in children: A comprehensive review
- Chapter 2: Temperament, executive functioning and anxiety in school-age children who stutter
- Chapter 3: Portuguese school-age children's experience of stuttering
- Chapter 4: Stuttering impact: A shared perception for parents and children?
- Chapter 5: How do teachers perceive the impact of stuttering on school-age children?

This dissertation ends with the discussion that encompasses all studies and the global conclusions.

All necessary appendices and annexes will be made available after listing bibliographical references. In order to comply with and protect copyright, the instruments used in this work will not be made available in their entirety.

CHAPTER 1: Stuttering in children: A comprehensive review[1]

[1] The following study is under review and awaiting for acceptance: Rocha, M.S., Yaruss J.S. & Rato J.R. (under review). Stuttering in children: A comprehensive review. *Communication Science & Disorders.*

Abstract

Stuttering is a fluency disorder in which the flow of speech is disrupted. The disorder is frequently misunderstood; to better analyze it is necessary to understand stuttering as more than a speech problem. It should instead be viewed as a communication disorder with the potential to affect several aspects of children's daily 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.

A comprehensive review of the literature was conducted to summarize and analyze previously published research considering stuttering as a dynamic disorder influenced by several factors. This review focuses on the development of this disorder, and the implications for the onset, manifestation, and chronicity of stuttering in school-age children who stutter.

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. This comprehensive view contributes to a better understanding of therapeutic and scientific factors that must be considered in the evaluation and treatment of stuttering.

1. The Stuttering Phenomenon

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 (Colton & Casper, 1996). 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 (American Speech-Language-Hearing Association, 1993, 2014; Yaruss, 1998; Yaruss & Quesal, 2004). 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 (Ambrose & Yairi, 1999).

Stuttering can be described as neurodevelopmental disorder (Smith & Weber, 2017) that normally arises in young children. 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 (Ambrose & Yairi, 1999; Guitar, 2014). The disfluencies associated with stuttering are commonly called stuttering-like disfluencies (Yairi, 2007). People who stutter often exhibit negative feelings, thoughts and attitudes toward their speech and experience adverse impact on quality of life (Yaruss & Quesal, 2004, 2016).

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 (Ambrose & Yairi, 1999; Tumanova, Conture, Lambert, & Walden, 2014).

Over the years, there have been attempts to define stuttering with a multidimensionality 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 (Travis, 1978). Also Johnson's research was an important contribution to the field of stuttering. Probably, Diagnosogenic theory (1938) stating that: stuttering onset was related with the overreaction from parents to child's disfluencies, was the basis for the more in-depth studies on the influence of the environment (Johnson, 1938). 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 (Choo, Burnham, Hicks, & Chang, 2016; Eichorn, Marton, & Pirutinsky, 2018; Leclercq, Suaire, & Moyse, 2018; Smith & Weber, 2017; Yaruss & Quesal, 2006). 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 (Johnson, 1958). Each blind man, who is examining the elephant, will come to different conclusions, as he is only examining a part of the animal (Manning, 2001).

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" (Sheehan, 1970). 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 (Andrews et al., 1983; Cooper & Cooper, 1985; De Nil, 1999; Neilson & Neilson, 1987; Riley & Riley, 1979; Smith & Kelly, 1997; Wall & Myers, 1984).

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 (Perkins, 1990). Current research on stuttering has supported the loss of control as part of how speakers experience stuttering (Tichenor & Yaruss, 2019).

Relatively recent, there have been efforts to understand stuttering in the light of 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 (WHO, 2001). 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) (WHO, 2001; Yaruss & Quesal, 2004; Tichenor & Yaruss, 2019).

The DSM-V definition of stuttering takes into account some of the functionality concepts mentioned above (Association American Psychiatrics, 2013). 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 reinforce 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 (Ambrose & Yairi, 1999; Reilly et al., 2013; Yairi & Ambrose, 2005). 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 (Bloodstein & Bernstein Ratner, 2008; Yairi & Ambrose, 2005). Stuttering tend to persist in children with specific risk factors that can also influence the beginning, maintenance, or severity of stuttering (Yairi & Ambrose, 2005; Yairi & Seery, 2010). Research on those risk factors is ongoing (e.g. Choo, Burnham, Hicks, & Chang, 2016; Erdemir, Walden, Jefferson, Choi, & Jones, 2018; Franken et al., 2018; Nandhini, Thalamuthu, Valarmathi, Karthikeyen, & Srisailapathy, 2018).

2. 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 evidence about the existence of a genetic predisposition. Research has identified genetic mutations associated with stuttering (Frigerio-Domingues et al., 2019; Kazemi, Estiar, Fazilaty, & Sakhinia, 2018; Watkins, Chesters, & Connally, 2016; Wittke-Thompson et al., 2007); however there are, as yet, no definitive results regarding transmission models, chromosomes, genes, or sex factors that are involved in genetic expression (Kraft & Yairi, 2011; Watkins et al., 2016). This genetic predisposition can be triggered by neurophysiological factors, environmental, temperament, and language development (Anderson, Pellowski, Conture, & Kelly, 2003; Guitar, 2014).

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 (Smith & Weber, 2017).

This view is consistent with the Dual Diathesis - Stressor model (DD-S) of developmental stuttering (Choi, Conture, Walden, Jones, & Kim, 2016; Walden, Frankel, Buhr, Johnson, & Karrass, 2012). 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 (e.g., Alm, 2014; Ambrose, Yairi, Loucks, Seery, & Throneburg, 2015; Conture, Kelly, & Walden, 2012; Eggers, De Nil, & Van den Bergh, 2013; Smith & Weber, 2017; Yaruss & Quesal, 2016).

Endogenous abilities play an important role in the development of stuttering (Choi, Conture, Walden, Jones, & Kim, 2016), and there is a growing scientific interest about how temperament (e.g. Eggers, De Nil & Van den Bergh, 2010) and executive functions (e.g. Costelloe et al., 2015) may influence this process. Therefore, these abilities can be thought of as a part of the *diathesis* that may contribute to the stuttering phenomenon (Choi et al., 2016).

Temperament studies have concluded that children who stutter are more likely to be reactive and sensitive compared to their nonstuttering peers (e.g. Ambrose et al., 2015), with a tendency for impulsivity (Costelloe, Cavenagh, & Davis, 2015; Eggers, De Nil, & Van den Bergh, 2010). Some studies have also reported that children who stutter have difficulty adapting to new objects and situations (e.g. Schwenk, Conture, & Walden, 2007), exhibit a greater negative effect (e.g. Ntourou, Conture, & Walden, 2013), and may have difficulties in self-regulation (e.g. Johnson, Walden, Conture, & Karrass, 2010). Temperament and EF have common strands; however, they have been investigated separately (Sudikoff, Bertolin, Lordo, & Kaufman, 2015). For example, certain temperamental characteristics, such as attentional focusing (Rothbart, Ahadi, & Evans, 2000), are assumed to have cognitive underpinnings in the executive or anterior attention system (Simonds, 2006; Sudikoff et al., 2015)

Previous studies reported that children who stutter are less successful in maintaining attention (Costelloe et al., 2015; Heitmann, Asbjørnsen, & Helland, 2004; Kaganovich, Wray, & Weber-Fox, 2010) and selecting information from sensory input (Eggers, De Nil, & Van den Bergh, 2012). Findings also indicate a tendency for impulsivity compared to nonstuttering peers (Bajaj, 2007; Katerina Ntourou, Anderson,

& Wagovich, 2018; Wolfe & Bell, 2004). In addition, studies indicate that children who stutter perform less well in working memory than their nonstuttering peers (Anderson & Wagovich, 2010; Oyoun, El Dessouky, Shohdi, & Fawzy, 2010). Reflecting on the close relationship between cognition and language, it is interesting to note prior studies reporting differences in children's speech sound (e.g. Anderson, Wagovich, & Hall, 2006) (Anderson, Pellowski, & Conture, 2005; Coulter, Anderson, & Conture, 2009; Rocha, Reis, & Carmona, 2016) and more advanced language skills (Millard, 2008).

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 (Craske et al., 2009; Daniels, Gabel, & Hughes, 2012; Walden et al., 2012). 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 (Eaves, Silberg, & Erkanli, 2003; Silberg, Rutter, Neale, & Eaves, 2001).

Anxiety has been associated with stuttering, yet its relationship with stuttering is still controversial (Craig, 2014; Iverach & Rapee, 2014; Manning & Beck, 2013). Although several reports indicate that adults who stutter experience elevated levels of anxiety (Alm & Risberg, 2007; Craig & Tran, 2014; Iverach, Menzies, Brian, Packman, & Onslow, 2011), people who stutter do not necessarily have to be anxious. They may experience anxiety in social situations involving speech, however (Alm, 2014; Iverach & Rapee, 2014; W. Manning & Beck, 2013; McAllister, Kelman, & Millard, 2015; Messenger, Packman, Onslow, Menzies, & O'Brian, 2015).

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 (Iverach et al., 2017; Iverach & Rapee, 2014; McAllister et al., 2015; Messenger et al., 2015; Mulcahy, Hennessey, Beilby, & Byrnes, 2008). Some other studies have found no specific trend toward elevated anxiety in children (Andrews & Harris, 1964; Craig & Hancock, 1996; Merwe, Robb, Lewis, & Ormond, 2011; Ortega & Ambrose, 2011).

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 activites and participation in society (Yaruss & Quesal, 2006).

3. 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 (Smith & Weber, 2017; Yaruss & Quesal, 2016).

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; Riley, 2009) 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 (e.g., Mancinelli, 2019; Manning & Gayle Beck, 2013; Sonneville-Koedoot, Stolk, Raat, Bouwmans-Frijters, & Franken, 2014).

The *Communication Attitude Test* (CAT; Brutten & Vanryckeghem, 2006) and the *Overall Assessment of the Speaker's Experience of Stuttering – Ages 7-12* (OASES-S; Yaruss & Quesal, 2016) 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 which includes 35 true/false statements about speech-associated attitudes of school-age children who stutter. Psychometric measures shows 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 (Bernardini, Vanryckeghem, Brutten, Cocco, & Zmarich, 2009; Brutten & Vanryckeghem, 2006; Guttormsen et al., 2015).

The OASES-S is another self-report instrument. It is based on the WHO's ICF as adapted to stuttering by Yaruss (1998) and Yaruss and Quesal (2004). 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 (Yaruss & Quesal, 2016), 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 (Beilby, Byrnes, & Yaruss, 2012; Blumgart, Tran, Yaruss, & Craig, 2012; Bodil, Sønsterud, & Kirmess, 2018; Chun, Mendes, Yaruss, & Quesal, 2010; Euler, Anders, & Merkel, 2016; Freud, Kichin-Brin, Ezrati-Vinacour, Roziner, & Amir, 2017; Koedoot, Bouwmans, Franken, Stolk, 2011; Lankman, Yaruss, & Franken, 2015; Osborn, Yaruss, Quesal, Schiefer, & Chiari, 2012; Rocha et al., 2019; Sakai, Chu, Mori, & Yaruss, 2017; Yadegari et al., 2018).

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* (Millard, Edwards, & Cook, 2009), and observational rating scales, such as those included in the *Test of Childhood Stuttering* (TOCS; Gilliam, Logan, & Pearson, 2009). Other forms designed to collect comprehensive history can be found in books and in treatment programs (e.g. Guitar, 2014; Cooper & Cooper, 1985; Haynes & Pindzola, 1998; Reardon- Reeves & Yaruss, 2017; Silverman, 2004; Woolf, 1967; Yaruss & Reardon- Reeves, 2017).

4. 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 (Boey, 2012; Diehl, Robb, Lewis, & Ormond, 2018; Guttormsen, Kefalianos, & Næss, 2015; Manning & Beck, 2013; Yaruss & Quesal, 2016).

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 (Craig, Blumgart, & Tran, 2009; Yaruss & Quesal, 2004).

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 (Cooper & Cooper, 1996; Enderby & Emerson, 1996; Hayhow, 1999; St. Louis et al., 2016). 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 (St. Louis et al., 2016). For example, findings of Valente and colleagues with Public Opinion Survey of Human Attributes-Stuttering (POSHA-S), revealed notable differences between countries and cultures across Europe (Valente, St. Louis, Leahy, Hall, & Jesus, 2017).

According to several reports, children are aware of their stuttering shortly after the onset. As they grow, the impact of the disorder may increase (Hollister & Hollister, 2015; Vanryckeghem, Hylebos, Brutten, & Peleman, 2001).

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 (Crichton-Smith, 2002; Daniels, Gabel, & Hughes, 2012; Hayhow, Cray, & Enderby, 2002; Klompas & Ross, 2004). 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 (Etchell, Civier, Ballard, & Sowman, 2018; Nederlandse Vereniging voor Logopedie en Foniatrie, 2014).

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 (Alm, 2014; Berquez & Kelman, 2018). 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 (Hugh-Jones &

Smith, 1999; Langevin, 2009). Langevin, Packman, and Onslow (2010) 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 and parents' view regarding the impact of stuttering (Guttormsen, Yaruss, & Næss, 2019; Rocha, Yaruss, & Rato, 2019).

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 (Alm, 2014; Rasskazov & Rasskazova, 2007). School-age children divide most of their daily time between home, and at school with their teachers and classmates (Boey, 2012; Seixas, Matos, Festas, & Fernandes, 2014), so the perspective of teachers is particular important.

Some reports highlight the negative perceptions and stereotypes held by teachers regarding people who stutter (Dorsey & Guenther, 2000; Turnbull, 2006). Other studies highlight a general lack of knowledge (Li & Arnold, 2015; Panico, Daniels, & Claflin, 2011; Silva et al., 2016). Such findings highlight the need to improve teamwork between professionals.

5. Conclusion

The misunderstandings that remain in the field of 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 which 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 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.

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CHAPTER 2: Temperament, executive functioning and anxiety in school-age children who stutter[2]

[2] The following study was reviewed by peers and accepted for publication: Rocha, M.S., Yaruss J.S. & Rato, J.R. (2019). Temperament, Executive Functioning, and Anxiety in School-Age Children Who Stutter. *Frontiers in Psychology*. doi: 10:10.3389/fpyg.2019.02244 (Annex 2)

Abstract

The purpose of this study was to examine temperament dimensions, executive functioning ability, and anxiety levels in school-age children who stutter and their nonstuttering peers. Participants were 100 Portuguese children aged 7 to 12 years (*M*=9.13; *SD*=1.70), including 50 children who stutter and 50 children who do not stutter. Analyses, which were performed separately for younger and older participants, sought to identify correlations between key variables. Temperament was evaluated through a parent questionnaire, executive functioning was evaluated through children's responses on a performance test, and anxiety level was assessed through a self-perception scale. On the temperament measure, comparisons between children who stutter and their nonstuttering peers revealed that older children who stutter exhibited significantly higher scores on the Anger/Frustration, Impulsivity, and Sadness subscales, and lower averages on the Attention/Focusing, Perceptual sensitivity, and Soothability/Falling reactivity subscales. On the executive functioning task, comparisons revealed that the group of younger children who stutter exhibited significantly higher average execution times than their nonstuttering peers. There were no statistically significant differences in anxiety between children who stutter and children who do not stutter, and there were no statistically significant correlations between temperament factors and measures of executive functioning. Children who stutter experienced lower ability to orient attention and greater emotional reactivity compared with their nonstuttering peers. Significant correlations were found between executive functioning and age and among the temperament factors themselves. These results, which support the need for a multidimensional view of stuttering, were interpreted in the context of the Dual Diathesis - Stressor model. Findings indicate that temperament and executive functioning abilities may contribute to the development of stuttering.

1. Introduction

1.1 Temperament

Temperament is an overarching term for a collection of traits that are assumed to be biologically determined and related to individual differences in reactivity and self-regulation (Jones et al., 2014; Rothbart et al., 2000).

Temperament can develop over time (Goldsmith et al., 1987) and be influenced by environmental interactions (Eggers et al., 2010). According to Rothbart and colleagues, "constitutional" factors are associated with genes and environment, "reactivity" is related to sensory response systems, and "self-regulation" relates to the process that can facilitate or inhibit reactivity (Rothbart et al., 2000). Thomas and Chess (1996) described nine temperament dimensions: "Activity Level", "Rhythmicity", "Approach/Withdrawal", "Adaptability", "Threshold of Responsiveness", "Intensity of Reaction", "Quality of Mood", "Distractibility", "Attention Span", and "Persistence". The authors relate temperament to the expression of a particular behavior. Children's and adults' intrinsic motivations and abilities for a specific behavior can be mediated by aspects of their temperament, such as their activity level, their adaptability, and their persistence (Goldsmith et al., 1987). Some authors have connected temperament differences in children who stutter with their susceptibility to begin, continue, or recover from stuttering (Ambrose et al., 2015; Conture, 2001; Guitar, 2014). Specifically, studies have suggested that children with a sensitive temperament may have neural vulnerabilities that cause them to be more likely to develop stuttering (Guitar, 2014).

Findings regarding temperament in children who stutter have been inconsistent. Therefore, it is not yet possible to draw firm conclusions about differences in temperament between children who stutter and their nonstuttering peers. Still, there is an increasing literature reporting a propensity for a more reactive and sensitive temperament in children who stutter (Alm, 2014; Ambrose et al., 2015; Eggers et al., 2010; Embrechts et al., 2000; Felsenfeld et al., 2000; Karras et al., 2006), and there is indication that more reactive and sensitive children tend to respond more strongly to disruptions in speech fluency (Walden et al., 2012).

Temperamental characteristics in preschool children that have been shown to contribute to stuttering include difficulty concentrating on tasks (Anderson et al., 2003; Embrechts et al., 2000), and low frustration tolerance (Eggers et al., 2010; Reilly et al., 2009; Druker et al., 2019). According to Spaulding et al. (2008), tasks dependent on sustained selective attention may be influenced by limited processing resources and situational demands. It is also known that attentional control plays an important role in children's ability to manage and regulate their emotions (Blair and Ursach, 2011). Several studies have reported that preschool children who stutter are prone to have difficulty adapting to new objects and situations (Anderson et al., 2003; Eggers et al.,

2010; Embrechts et al., 2000; Hollister, 2015; Howell et al, 2004; Reilly et al., 2009; Schwenk et al.,2007) and have a tendency toward greater negative affect (Ntourou et al., 2013; Embrechts et al., 2000) and negative mood (Howell et al., 2004). Experimental studies of the temperament of preschool children who stutter have revealed a tendency for impulsivity (Eggers et al., 2010; Schwenk et al., 2007) and for lower self-regulation, or the ability to regulate emotional behaviors (Ntourou et al., 2013; Johnson et al., 2010).

While studies of temperament in preschool children and adults who stutter have revealed notable differences compared to peer groups who do not stutter (e.g. Ambrose et al., 2015; Ntourou et al., 2013; Reiley et al., 2009; Smith and Weber, 2017), temperament studies involving school-age children are more rare(Nicholas et al., 2015; Oyler, 1996). Those that have been conducted have shown that children of this age who stutter tend to be more sensitive and withdrawn than their nonstuttering peers (Fowlie and Cooper, 1978). There is a need to further research temperament in school-age children in order to understand the changes that arise throughout a child's development. In the same way that some studies conclude that young children and adults who stutter exhibit certain temperament characteristics, it is important to determine whether these characteristics maintain or otherwise change during the school age and how they contribute to cognitive development (Singer and Fagen, 1992).

1.2 Executive Functioning

The role of EF in childhood stuttering has been a subject of increased attention in recent years (Jones et al., 2014; Ntourou et al., 2013). EF is a term used to describe a diverse set of cognitive skills needed to perform activities that require planning and monitoring of intentional behaviors that allow individuals to interact with the world in an adaptive and appropriate way (Diamond, 2013). Researchers have highlighted three basic components of EF: inhibition, the ability to suppress a prepotent response; working memory, which implies an information updating process; and shifting, the ability to shift between tasks or mental sets and is an important aspect of executive control (Miyake et al., 2000). Despite some inconsistencies in findings across studies, several studies have shown that children who stutter, especially in earlier ages, have a tendency to be less successful in maintaining attention than their typically fluent peers (Costelloe et al., 2015; Eichorn et al., 2017; Heitmann et al., 2004; Kaganovich et al.,

2010). Children who stutter are also prone to be less able to select information from sensory input (Eggers et al., 2012), more likely to exhibit impulsivity (Eggers et al., 2013), and more likely to have greater concern about their performance (Eichorn et al., 2017).

There seems to be symptoms, similar to attention deficit disorders, for some children who stutter (Anderson et al., 2003; Druker et al., 2019); however, studies related to the incidence of attention deficit disorders are not conclusive and are performed with a limited sample size (Donaher and Richels, 2012; Riley and Riley, 2000). Children who stutter tend to perform less well than their peers in working memory (Anderson and Wagovich, 2010; Oyoun et al., 2010), inhibitory control (stroop-like tasks), and attentional focusing, as indicated through parenting ratings (Bajaj, 2007; Wolfe and Bell, 2004). Difficulties related to inhibitory control and attentional focusing are especially evident in studies that use parent-report questionnaires (Ofoe et al., 2018).

Cognitive processes described above are closely linked to emotional regulation (Sudikoff et al., 2015) and can influence the experience of anxiety (Craske et al., 2009).

1.3 Anxiety

Anxiety is a general term for an individual's emotional struggle that combines nervousness, fear, apprehension, and worrying (Craske et al., 2009). According to some authors (e.g., Craig and Hancock, 1996; Craig et al., 2003; Craig and Tran, 2014; Ezrati and Levin, 2004), anxiety can be divided into *trait* anxiety (related to stable anxious baseline characteristics) and *state* anxiety (related to transitory conditions due to unpleasant emotional arousal with a tendency to appear when people have to cope with demanding situations). People who stutter often struggle with *state anxiety*, since anxiety will likely become a secondary effect of living with stuttering condition rather than being a static condition (Alm and Risberg, 2007; Messenger et al., 2015). Also, according to Samochiş et al. (2011), increased anxiety is a normal reaction to the physical aspects of stuttering. Nevertheless, some studies have not supported a relationship between anxiety and stuttering or have found little significant differences (e.g. Andrews and Harris, 1964; Andrews et al., 1983; Cox et al., 1984; Craig and Hancock, 1996; Hedge, 1972; Peters and Hulstijin, 1984).Currently, the occurrence of anxiety in children who stutter is still a subject of debate (Alm, 2014; Alm and Risberg,

2007; Craig, 2014; Manning and Beck, 2013; Smith et al, 2014). Even in the literature that does support the existence of anxiety in children, the age at which anxiety symptoms begin to appear has not yet been identified. Specifically, the studies linking anxiety to preschool-age have shown no differences between children who stutter and nonstuttering peers on anxiety measures and salivary cortisol levels (Van der Merwe et al., 2011).Some studies have found significantly higher anxiety symptoms in school age children who stutter, ages 7 to 12 (e.g. Iverach et al., 2011), and other studies have reported the same for children from 10 and up (Davis et al., 2007; Iverach et al., 2017; McAllister et al., 2015; Mulcahy et al., 2008). Nevertheless, other studies have not found any trend toward elevated anxiety in school age children (Andrews and Harris, 1964; Craig and Hancock, 1996; Ortega and Ambrose, 2011) . Some evidence suggests that the levels of anxiety tend to increase over time and can exceed normal values in adolescence and adulthood (Mulcahy et al., 2008). Still, the meaning of these findings is unclear and according to Messenger and colleagues (2015), adolescents who stutter may try to present themselves positively to hide their true concerns about stuttering. This lack of consistency suggests the existence of other variables that might affect the development of anxiety.

1.4. Temperament, EF, Anxiety and the Dual diathesis-stressor model

To date, no studies have simultaneously considered the relationship between temperament, EF, and anxiety in children who stutter, even though all of these factors are believed to affect children who stutter. Because of the relationship between anxiety, temperament, and EF (Nigg, 2000), considering these factors in concert will help to elucidate how these issues relate to the development and experience of stuttering.

There is already a large body of empirical evidence suggesting a strong concurrent relationship between temperament characteristics and executive functioning (EF) (Simonds, 2006; Sudikoff etal., 2015). According to Affrunti and colleagues (2015) the expression of temperament may be influenced by executive functioning. Temperament also includes behavioral aspects, such as approach and withdrawal, as well as attentional processes, including orientation maintenance and executive control. Together, these abilities are the building blocks of the development of self-regulation (Rothbart and Hwang, 2002). Studies of cognitive development have shown that attention control, inhibition of inappropriate behavior, decision making, and other

cognitive processes that occur in emotionally demanding contexts, are strongly supported by EF (Gupta et al., 2011).

Research has further identified temperamental characteristics and cognitive abilities as predictors of anxiety (Kefalianos et al., 2012). Environmental factors can be part of these dynamic interactions and, together with temperamental characteristic and cognitive abilities, influence how children deal with stuttering. Because temperament characteristics and EF abilities may contribute to a child's likelihood of responding to experiences in a particular way, the involvement of temperament and EF in the development of stuttering can be described in terms of the dual diathesis-stressor (DD-S) model (Walden et al., 2012). The DD-S model proposes that endogenous abilities of children who stutter interact in a dynamic way with exogenous contexts (stressors). In line with this model, temperament and EF characteristics can be seen as a diathesis that can be triggered by a stressor, transforming a predisposition to an actual emotional response in a particular situation. As applied to stuttering, the theory suggests that a child's endogenous characteristics related to temperament, anxiety, and EF, may be affected by exogenous stressors that may increase (or decrease) the frequency of stuttering. Importantly, exogenous contexts (stressors) can activate cognitive and affective processes and pushing the autonomic nervous system out of homeostasis, thereby increasing the emotional response (Walden et al., 2012). This imbalance can translate into anxiety and other signs of dysregulation (Craske et al., 2009).

The present study intended to address the literature gap on the research of temperament, EF, and anxiety jointly, comparing school-age children who stutter and nonstuttering peers. The combination of these three aspects can give us further information about the interaction between emotional and cognitive factors. Moreover, the DD-S model, which focuses the interaction between intrinsic and external factors and how they may change over time, highlights the need to concurrently consider factors such as temperament, EF, and anxiety. Taken together, these factors can provide more clues about the onset, development, and possible persistence of stuttering during childhood. A better understanding of such relationships may help clinicians better understand how stuttering affects children and this understanding may contribute to the development of more effective and personalized treatment programs.

2. Methods

2.1 Participants

Participants were 100 Portuguese children, 50 children who stutter ("S" Group) and 50 age-matched children who do not stutter ("N" Group), ages 7 to 12 years old. The Stuttering Severity Instrument – 4th Edition (SSI-4) (Riley, 2009) was used to confirm and diagnose stuttering.

Table 1 shows the demographic characteristics of the participants. The sex ratio of participants who stutter was 2.6 males to each female; for participants who do not stutter, it was 0.8 males to each female. This sex ratio for children who stutter is consistent with previous literature (Yairi and Ambrose, 2005; Craig et al., 2002).

In order to explore developmental differences, the participants who stutter (n=50) and their nonstuttering peers (n=50) were grouped according to age: younger children (7-9 years old; M=7.92; SD=0.81) and older children (10-12 years old; M=10.95; SD= 0.82). The cutoff age point for the 2 groups in this study was based on the development and important changes that take place during this period, where previously acquired learning is consolidated and new intellectual, psychological and social acquisitions arise (Blake and Pope, 2008). In addition, this age group distinction corresponds to the first two education cycles in Portugal: the first cycle includes the first 4 years of school (about 7-9 years old) and the second cycle includes the 5th and 6th grades (about 10-12 years old). Depending upon a child's birth date, however, it is possible to find children in the 7th grade who are 12 years old. Pre-school education in Portugal is intended for children between 3 and 6 years old; from the age of 13, Portuguese children are usually in high school (Alarcão et al., 2009).

Inclusion and exclusion criteria ensured that children did not exhibit any neurological or psychiatric impairment, learning disorder, or history of head injury or seizures. The sample was chosen by convenience: participants who stutter were recruited from speech-language therapists and through referral of school teachers; participants who do not stutter were recruited in some schools attended by their stuttering peers. All children were monolingual speakers of Portuguese.

When the study was performed, 22% of the children who stutter were in speech therapy, 22% had previous speech therapy, and 28% were waiting for therapy or just initiating speech therapy. The children who were in therapy at the time of data collection had been in treatment between 1 month to 96 months (M= 9.30 mos.; SD =

19.38 mos.). Children who had previous therapy had received between 3 months and 48 months of treatment (M=13.28 mos.; SD = 12.99 mos.).

Table 1: Demographic characteristics of the participants (children who stutter=50; children who do not stutter =50)

Group	Children who stutter	Children who do not stutter	Total
Age mean (SD)	9.10 (1.73)	9.16 (1.68)	9.13 (1.70)
Sex (M/F)	36/14 (72%/28%)	22/28 (44%/56%)	58/42 (58%/42%)
Education Level (n)			
1st Grade	8 (16%)	3 (6 %)	11 (11%)
2nd grade	11 (22%)	10 (20%)	21 (21%)
3rd grade	9 (18%)	8 (16%)	17 (17%)
4th grade	7 (14%)	14 (28%)	21(21%)
5th to 7th grade	15 (30%)	15 (30%)	30 (30%)
Treatment (n)			
Without treatment	14 (28%)	_	_
Speech Therapy	11 (22%)	_	_
Waiting or initiating	14 (28%)	_	_
Previous Therapy	11 (22%)	_	_

2.2. Materials

The SSI-4 (Riley, 2009) was used along with the Portuguese story, "*A história do rato Artur*" (Guimarães, 2007), "Rato Artur" story has been used in several Portuguese studies (e.g. Guimarães and Abberton, 2005; Silvestre, 2009; Silvestre et al, 2011) because it has a high test-retest consistency and is phonetically balanced. This has being interpreted that is close to spontaneous discourse (Moon et al., 2012). Eight of the 7-year-old participants had difficulties reading the story, so only the SSI-4 plates were used for those participants.

The parents provided information about socio-demographic background, and the child's stuttering via a checklist created for this study. Table 1 shows information about children; table 2 shows information about parents' sex, age, education level, and family history of stuttering.

Table 2: Demographic characteristics of parents (parents of children who stutter=50; parents of children who do not stutter =50)

Group	Parents of children who stutter		Parents of children who do not stutter		Total	
Age mean (SD)	42.26 (4.82)		39.60 (4.34)		40.93 (4.76)	
Sex (M/F)	6/44 (6%/88%)		3/47 (6%/94%)		9/91/ (9%/91%)	
Family history of stuttering (n)						
Yes	30 (60%)		___		___	
No	20 (40%)		___		___	
Education Level (n)	Mother	Father	Mother	Father	Mother	Father
1-4 years	0 (0%)	0 (0%)	0 (0%)	2 (4%)	0 (0%)	2 (2%)
5-6 years	0 (0%)	0 (0%)	2 (4%)	0 (0%)	2 (2%)	0 (0%)
7-9 years	5 (10%)	6 (12 %)	2 (4 %)	7 (14%)	7 (7%)	13 (13%)
10-12 years	8 (16%)	10 (20%)	15 (30%)	19 (38%)	23 (23%)	29 (29%)
Graduation	32 (64%)	30 (60%)	27 (54%)	21 (42%)	59 (59%)	51 (51%)
Master	3 (6%)	4 (8%)	3 (6%)	0 (0%)	6 (6%)	4 (4%)
Phd	2 (4%)	0 (0%)	1 (2%)	1 (2%)	3 (3%)	1 (1%)

2.2.1 Temperament

The Temperament in Middle Childhood Questionnaire (TMCQ) (Simonds and Rothbart, 2004) is a parent reported, paper and pencil measure that evaluates temperament in middle childhood (7-10 years old). It consists of 157 questions that examine 17 dimensions of temperament: 1) Activity Level, 2) Affiliation, 3) Anger/Frustration, 4) Assertiveness/Dominance, 5) Attention Focusing, 6) Discomfort; 7) Fantasy/Openness, 8) Fear, 9) High Intensity Pleasure, 10) Impulsivity, 11) Inhibitory Control, 12) Low Intensity Pleasure, 13) Perceptual Sensitivity, 14) Sadness, 15) Shyness, 16) Soothability/Falling reactivity, 17) Activation Control (see Table 3). Answers are obtained by parents rating their children on a 5-point Likert scale ranging from “Almost always untrue” to “Almost always true,” with the option of “Does not apply.”

Through the TMCQ, it is possible to identify reactivity/sensitivity and self-regulation characteristics. For example, the TMCQ scales such as Anger/frustration are connected to reactivity, whereas scales such as Inhibitory control are more related to self-regulation (Eggers et al., 2013). For example, young children may become angry and impulsive when their goals are hindered. This might occur when they have to wait for something they want (Rothbart et al, 2001).

Of the 17 dimensions of temperament that are part of the instrument, 13 dimensions derive from the well-validated Children's Behavior Questionnaire (CBQ: Rothbart et al, 2001), which has been used in several studies to investigate the relationship between temperament and stuttering (e.g. Ambrose et al., 2015; Eggers et al., 2010). In Simonds (2006), the TMCQ was shown to have good internal consistency reliability (Cronbach's alpha ranged from 0.62 to 0.83) and acceptable agreement between self-report and parent report (Pearson's r ranged from -0.02 to 0.50).

The questionnaire was translated to European Portuguese for this study (Rocha and Rato, 2017).

Table 3: TMCQ scale (Simonds and Rothbart, 2004) descriptions and sample items

TMCQ Scale	Definition
Activity Level	Level of gross motor activity including rate and extent of locomotion.
Affiliation	The desire for warmth and closeness with others, independent of shyness or extraversion.
Anger/Frustration	Amount of negative affect related to interruption of ongoing tasks or goal blocking.
Assertiveness/Dominance	Tendency to speak without hesitation and to gain and maintain control of social situations
Attentional Focusing	Tendency to maintain attentional focus upon task-related channels.
Discomfort	Amount of negative affect related to sensory qualities of stimulation, including intensity, rate or complexity of light, movement, sound, and texture.
Fantasy/Openness	Active imagination, aesthetic sensitivity, intellectual curiosity.
Fear	Amount of negative affect, including unease, worry or nervousness related to anticipated pain or distress and/or potentially threatening situations.
High Intensity Pleasure	Amount of pleasure or enjoyment related to situations involving high stimulus intensity, rate, complexity, novelty, and incongruity.
Impulsivity	Speed of response initiation.
Inhibitory Control	The capacity to plan and to suppress inappropriate approach responses under instructions or in novel or uncertain situations.
Low Intensity Pleasure	Amount of pleasure or enjoyment related to situations involving low stimulus intensity, rate, complexity, novelty, and incongruity
Perceptual Sensitivity	Amount of detection of slight, low intensity stimuli from the external environment.
Sadness	Amount of negative affect and lowered mood and energy related to exposure to suffering, disappointment, and object loss.
Shyness	Slow or inhibited approach in situations involving novelty or uncertainty.
Soothability/Falling Reactivity	Rate of recovery from peak distress, excitement, or general arousal.
Activation Control	The capacity to perform an action when there is a strong tendency to avoid it.

2.3 Executive Functioning

Children were assessed using the Portuguese version of the Children's Color Trails Test (CCTT), a neuropsychological paper and pencil test of EF (Pinto, 2008). The CCTT measures sustained visual attention, sequencing, psychomotor speed, and cognitive flexibility. It is intended for ages 8 to 16, though the authors have reported success with children as young as 7 years old (Llorente et al., 2003). The test includes two parts (CCTT-1 and CCTT-2), each involving one trial and one experimental task. In CCTT1, the child must connect the numbers from 1 to 25 following a correct sequence as quickly as possible. In CCTT2, the child must repeat the task from CCTT1 but with a

color alternation. In this task, the child still connects the numbers from 1 to 25. This time, however, each number is repeated in different colors (i.e., there are yellow numbers and pink numbers), and the child must be sure to follow the numerical order even when it changes between yellow and pink (Llorente et al., 2003).

The results of both parts of this test consist of: a) time (in seconds) that the child takes to complete the tasks, b) the number of times almost failed (the failures), c) the number of errors, and d) the number of warnings (when a child makes a mistake, the examiner advises him or her to start the test again from the last correct circle).

CCTT has been increasingly used around the world (e.g., Llorente et al, 2009; Konstantopoulos et al, 2015; Koo and Min, 2008; Pinto, 2008) for the assessment of children in neurological and psychiatric disorders such as language disabilities (e.g. Williams et al., 1995), attention deficit/hyperactivity disorder (e.g., Cho et al., 2011; Kennel et al., 2010), and other conditions (Llorente et al, 2003). CCTT is based on the Trail Making Test, which assess speeded visuomotor tracking. Research has shown discriminant validity and sensitivity across cultures (Williams et al, 1995). The CCTT is expected to have the same validity as the Trail Making in the assessment of children with several disorders (Williams et al, 1995). In a study with 70 children diagnosed with attention deficit and hyperactivity disorder, CCTT exhibited appropriate test-retest reliability (Llorente et al, 2009).

2.4 Anxiety

The children also completed the Portuguese version of Multidimensional Anxiety Scale for Children (MASC), which examines the symptoms of anxiety in children and adolescents ages 7 to 19 years. It contains 39 questions, with 4-point Likert scale responses (Matos, et al., 2012; March et al., 1997; Salvador et al., 2017). Items on this questionnaire are grouped into 4 factors: a) Physical symptoms, b) Social anxiety, c) Separation anxiety, and d) Harm avoidance (Wei et al., 2014). Participants are asked to score statements such as: "I get nervous if I have to do something in public," choosing between: a) "it is never or almost never true," b) "it is rarely true," c) "sometimes it is true," and d) "It is often true."

The normative data for the MASC show that it is oriented mainly toward inherent characteristics (*trait* anxiety), though it is also influenced by transitory conditions and situations (*state* anxiety) (March et al, 1997). Decades of research confirm the robust features of the MASC. Several studies with general populations and

with clinical populations supported their internal consistency, temporal stability, and convergent validity (Salvador et al., 2017). The original English version demonstrated good internal consistency (between .60 and .90), strong convergent/divergent validity, and test-retest reliability (March et al., 1997). The Portuguese version of the MASC has also been shown to be an adequate and reliable measure for self-assessment of anxious symptomatology, presenting reasonable psychometric characteristics in internal consistency, temporal stability, and validity (Salvador et al., 2017).

2.3 Procedures

This study received full approval by the Ethics Committee of the Institute of Health Sciences of Universidade Católica Portuguesa (register number 34/2017). Prior to their participation in this study, parents signed a written informed consent for themselves and their children. Consent also included permission for the researcher to record the child and the right for participants to withdraw from the study at any time was clarified.

Children were assessed while parents completed the questionnaires. This was carried out in two sessions of approximately 30 minutes each.

All testing was conducted between December 2017 and May 2018. The SSI, MASC and CCTT instruments were applied on different days and in a different order, to reduce potential order effects that might bias results.

2.3.1 Temperament

Temperament was assessed using the Portuguese version of the TMCQ, with the 157 original questions, distributed in 17 temperament dimensions (Simonds and Rothbart, 2004). After a brief explanation from the researcher, parents completed the TMCQ. This required approximately 20 minutes. In addition to researcher's explanation, on the first page of the questionnaire parents could read instructions about the content of the questions and how to complete the form. After parents completed the questionnaire, the researcher scored the instrument according to the instructions.

2.3.2 Executive Functioning

For the EF assessment, the researcher presented and explained to the children how to perform the CCTT1, using the trial test. In both trial test and experimental test, children drew a line between the circles following a numerical order, as fast as they could; however, the CCTT1 trial test was performed with just 8 numbers. For the

CCTT2 the procedures were similar, with the difference that children should switched between colors (after a yellow circle the child should drew a line towards a pink circle, following a numerical order). The researcher recorded 9 scores for each child. These scores corresponded to: the time that the child took to complete the tests for both CCTT1 and CCTT2, as well as the number of warnings, failures, and wrong answers (Number Sequencing and Color Sequencing) (Llorente et al., 2003).

2.3.3 Anxiety

For the anxiety assessment, the MASC questionnaire was presented to each child. Children were asked to read all the questions and to choose the best option for each. Children were informed about the importance of responding to all questions. For 7-years-old children, the MASC questions were read in full by the examiner.

After the children completed the questionnaire, the researcher summed the items for each factor, obtaining 4 final scores, corresponding to: a) Physical symptoms, b) Social anxiety, c) Separation anxiety, and d) Harm avoidance, for each child.

2.4 Data Analysis

Preliminary analyses were made in order to check the assumptions of homogeneity. Results for some variables were not normally distributed; however, with the $n = 50$ for each participant group, the central limit theorem suggests that parametric tests (t-test) would still be sufficiently robust to avoid deviations from normality. Two-sample t-tests were used to compare mean scores for the stuttering and nonstuttering groups for the temperament (TMCQ), EF (CCTT), and Anxiety (MASC) measures. These analyses were performed separately for younger and older participants. A multivariate analysis using Principal Component Analysis (PCA) was performed in order to determine which variables were correlated and to summarize children characteristics in an ordination diagram. For the PCA analyses, younger and older children were grouped. This was done because of apparent differences between age groups. The use of PCA provided a dynamic view of the interaction among all of the variables, including age. To account for the large number of variables in the study (temperament, EF, anxiety and age) only the variables that showed statistical significance in the t-tests were used in the PCA. Data analysis was completed using SPSS (Statistical Package for the Social Sciences - Version 24 for windows, Armonk, NY: IBM Corp).

3. Results

3.1 Younger children group

3.1.1 Temperament

No statistically significant differences were found between groups of children who stutter and their nonstuttering peers ($p > .05$) for any of the variables of temperament including: 1) Activity Level, 2) Affiliation, 3) Anger/Frustration, 4) Assertiveness/Dominance, 5) Attention Focusing, 6) Discomfort; 7) Fantasy/Openness, 8) Fear, 9) High Intensity Pleasure, 10) Impulsivity, 11) Inhibitory Control, 12) Low Intensity Pleasure, 13) Perceptual Sensitivity, 14) Sadness, 15) Shyness, 16) Soothability/Falling reactivity, 17) Activation Control.

3.1.2 Executive Functioning

Group comparisons of the CCTT1 and the CCTT2 revealed that children who stutter exhibited significantly higher scores for execution time (CCTT1: $t_{(48.75)} = 3.144$, $p = .003$; CCTT2: $t_{(52.27)} = 3.753$, $p < .001$), as well as number of failures (CCTT1: $t_{(38.23)} = 2.627$, $p = .012$), number of warnings (CCTT1: $t_{(52.47)} = 2.968$, $p = .005$; CCTT2: $t_{(53.71)} = 3.757$, $p < .001$), number of sequencing errors (CCTT2: $t_{(34.99)} = 3.337$, $p = .002$), and color sequencing errors (CCTT2: $t_{(49.31)} = 2.416$, $p = .020$)(Table 4).

3.1.3. Anxiety

No statistically significant differences were found between groups of children who stutter and their nonstuttering peers ($p > .05$) for any of the variables of anxiety including: 1) Physical symptoms, 2) Social anxiety, 3) Separation anxiety, and 4) Harm avoidance, for each child.

Table 4: Mean (M), standard deviations (SD) and p-values for the temperament, EF and anxiety performance tasks for group of younger children who stutter (n=31; sex: M=25; F=6) and who do not stutter (n=31; sex: M=15; F=16)

	Children who stutter		Children who do not stutter			
Scores	*M*	*SD*	*M*	*SD*	*t*	*p*
Activation control	3.185	.442	3.326	.493	-1.183	.242
Activity level	3.632	.755	3.794	.709	-.087	.386
Affiliation	4.042	.361	4.033	.488	.086	.932
Anger/Frustration	3.251	.742	3.137	.552	.687	.994
Assertiveness/Dominance	3.122	.646	3.300	.597	-1.124	.265
Attention/Focusing	2.840	.993	3.513	1.990	-1.686	.097
Discomfort	2.819	.669	2.481	.669	1.993	.051
Fantasy/Openness	3.766	.660	3.857	.552	.586	.560
Fear	2.804	.689	2.612	.606	1.167	.298
High intensity pleasure	3.058	.651	2.998	.624	.373	.711
Impulsivity	2.983	.544	2.959	.523	.184	.854
Inhibitory control	2.962	.575	3.110	.587	-1.000	.322
Low intensity pleasure	3.256	.655	3.477	.629	-1.359	.179
Perceptual sensitivity	3.091	.835	3.206	.692	-.591	.557
Sadness	2.700	.452	2.713	.586	-.095	.925
Shyness	2.792	.811	2.651	.852	.664	.509
Soothability/Falling react.	3.223	.721	3.367	.571	-.831	.410
CCTT1 Time (sec)	86.308	33.943	64.032	20.107	3.144	**.003****
CCTT1 Number seq. errors	.193	.543	.069	.359	1.068	.290
CCTT1 Failures	.548	.961	.065	.359	2.627	**.012***
CCTT1 Warnings	1.677	1.558	.677	1.045	2.968	**.005****
CCTT2 Time (sec)	161.420	46.582	123.677	31.061	3.753	**<.001*****
CCTT2 Color seq. errors	1.355	1.279	.709	.772	2.406	**.020***
CCTT2 Number seq. errors	.419	.620	.032	.120	3.337	**.002****
CCTT2 Failures	1.452	1.480	.810	1.167	1.906	.061
CCTT2 Warnings	2.710	2.036	1.032	1.426	3.757	**<.001*****
Physical symptoms	6.258	4.885	7.267	5.836	.733	.467
Social anxiety	10.710	8.038	9.833	5.522	..498	.621
Separation anxiety	9.000	4.219	9.500	4.276	-.460	.647
Harm avoidance	19.774	4.566	19.700	4.276	.065	.948
Total score anxiety	45.420	15.000	46.267	14.694	-2.223	.824

*p< .05, **p < .01, ***p < .001

3.2. Older children group

3.2.1 Temperament

Statistically significant differences were found for several temperament factors (Table 5). Children who stutter scored lower than nonstuttering peers in Attention/Focusing ($t_{(36)}$=-3.526, p = .001), Perceptual Sensitivity ($t_{(36)}$= -2.411, p= .021), and Soothability/Falling reactivity ($t_{(36)}$= -2.932, p=.006). Children who stutter

scored higher than nonstuttering peers in temperament factors of Anger/Frustration ($t_{(36)} = 2.801$, $p = .008$), Impulsivity ($t_{(36)} = 2.899$, $p = .006$) and Sadness ($t_{(36)} = 3.683$, $p = .001$).

3.2.2 Executive Functioning

No statistically significant differences were found between groups of children who stutter and their nonstuttering peers ($p > .05$) for any of the variables of EF, including: 1) CCTT1 execution time, 2) CCTT1 number of sequencing errors, 3) CCTT1 number of failures, 4) CCTT1 number of warnings, 5) CCTT2 execution time, 6) CCTT2 number of color sequencing errors, 7) CCTT2 number of sequencing errors, 8) CCTT2 number of failures, 9) CCTT2 number of warnings.

3.2.3 Anxiety

As in the younger group, no statistically significant differences were found between groups of children who stutter and their nonstuttering peers ($p > .05$) for any of the variables of anxiety, including: 1) Physical symptoms, 2) Social anxiety, 3) Separation anxiety, and 4) Harm avoidance.

Table 5: Mean (M), standard deviations (SD) and p-values for the temperament, EF and anxiety performance tasks for group of older children who stutter (n=19 ; sex: M=11; F=8) and children who do not stutter (n=19 ; sex: M=7; F=12).

	Children who stutter		Children who do not stutter			
Scores	*M*	*SD*	*M*	*SD*	*t*	*p*
Activation control	3.049	.467	3.221	.406	-1.205	.236
Activity level	3.872	.694	3.806	.800	.271	.788
Affiliation	4.126	.449	4.171	.412	.320	.751
Anger/Frustration	3.335	.614	2.807	.546	2.801	**.008****
Assertiveness/Dominance	3.243	.736	3.324	.566	-3.81	.071
Attention/Focusing	2.644	.644	3.552	.920	-3.526	**.001*****
Discomfort	2.779	.627	2.584	.553	1.015	.317
Fantasy/Openness	2.916	.814	3.840	.536	-1.269	.212
Fear	2.916	.814	2.700	6.690	.881	.384
High intensity pleasure	3.084	.576	2.783	.655	1.501	.142
Impulsivity	3.084	.446	2.560	.650	2.899	**.006****
Inhibitory control	3.317	.448	3.536	.574	-1.312	.198
Low intensity pleasure	3.264	.476	3.435	.572	-.997	.325
Perceptual sensitivity	3.307	.567	3.722	.491	-2.411	**.021***
Sadness	3.036	.553	2.415	.485	3.683	**.001*****
Shyness	3.042	.986	2.838	.858	.679	.501
Soothability/falling reactivity	3.157	.402	3.663	.636	-2.932	**.006****
CCTT1 Time	51.745	15.000	52.790	19.472	-.185	.854
CCTT1 Number seq. errors	.263	.561	.211	.535	.296	.769
CCTT1 Failures	.158	.375	.000	.000	1.837	.074
CCTT1 Warnings	.632	1.065	.211	.419	1.604	.118
CCTT2 Times	97.9474	32.732	98.947	33.311	-0.093	.926
CCTT2 Color seq. errors	.579	1.610	.421	.838	.379	.707
CCTT2 Number seq. errors	.000	.000	.000	.000	.073	.943
CCTT2 Failures	1.105	1.370	.579	.837	1.429	.162
CCTT2 Warnings	.421	.961	.368	.831	.181	.858
Physical symptoms	8.842	7.654	6.800	3.721	1.051	.303
Social anxiety	10.263	7.001	11.526	4.937	.642	.525
Separation anxiety	9.474	6.040	8.526	4.033	.569	.573
Harm avoidance	17.947	4.972	18.158	4.375	-10.139	.891
Total score anxiety	46.579	19.585	44.158	10.569	.474	.638

*p< .05, **p < .01, ***p < .001

3.3 Multivariate analysis

The PCA ordination biplot (Figure 1) showed that CCTT2 Time (component loading=.82), CCTT2 warnings (component loading=.80), CCTT1 time (component loading= .75), age (component loading=-.69), CCTT2 number of sequencing errors (component loading= .67), CCTT1 warnings (component loading=.65), and CCTT2 Color sequencing errors (component loading=.51), were the variables influencing the

children's ordination along the first axis (Dimension 1), that is, the EF dimensions (Figure 1).

The right side of the axis shows the children with higher values of CCTT2 Time, CCTT2 warnings, CCTT1 time, CCTT2 number of sequencing errors, CCTT1 warnings, CCTT2 Color sequencing errors, and younger children. The left side of the axis shows children characterized by lower values of CCTT2 Time, CCTT2 warnings, CCTT1 time, CCTT2 number of sequencing errors, CCTT1 warnings, CCTT2 Color sequencing errors, and older children. Most of the children who stutter ("S") were plotted on the right side of the first dimension. The first axis accounted for 27.30% of the total variance. The parameters with greater contribution to the second axis (dimension 2 – Temperament dimensions) were Sadness (component loading= .78), Anger/Frustration (component loading= .75), Soothability/Falling reactivity (component loading= -.72), Attention/Focusing (component loading = -.69), and Impulsivity (component loading= .60). Most of the children who stutter were displayed on the upper part of the diagram, as they exhibited higher values of Sadness, Anger/Frustration and Impulsivity, and lower values of Soothability/Falling Reactivity and Attention/Focusing. The bottom part of the diagram shows mainly children who do not stutter, due to lower values of Sadness, Anger/Frustration and Impulsivity; and higher values of Soothability/Falling Reactivity and Attention/Focusing.

The second axis accounted for 20.04% of the total variance. The CCTT1 Time, CCTT1 Warnings, CCTT1 failures, CCTT2 Time, CCTT2 Warnings, CCTT2 number of sequencing errors, and CCTT2 color sequencing errors were highly and positively correlated with one another and negatively correlated with age. Sadness, Anger/Frustration, and Impulsivity were highly and positively correlated with each other; Attention, Soothability/Falling Reactivity and Perceptual Sensitivity were negatively correlated with Anger/Frustration, Impulsivity, and Sadness.

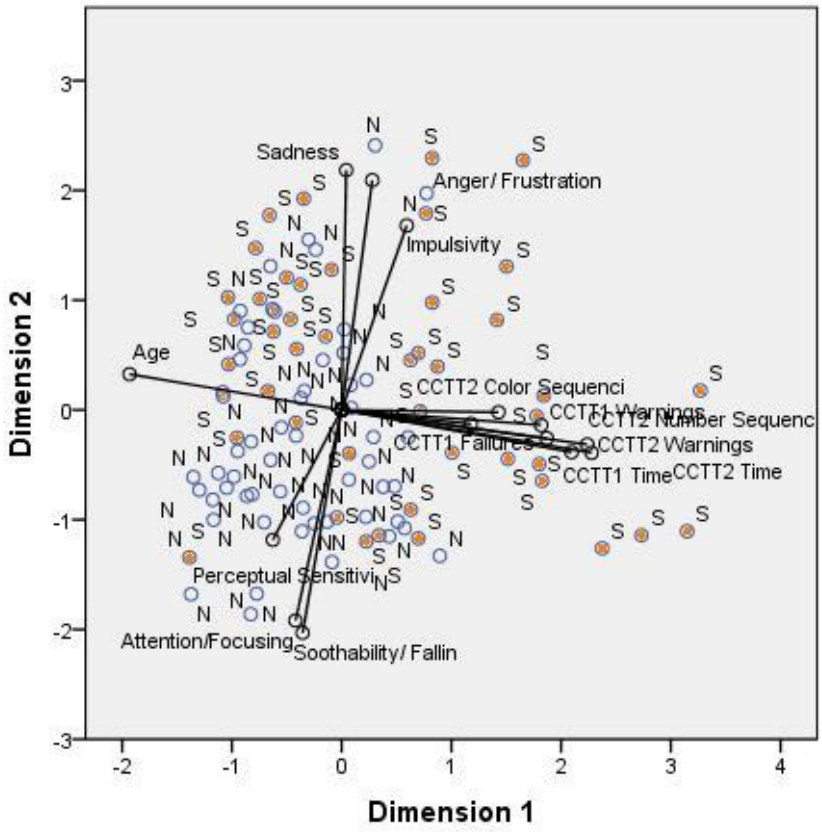

Figure 1: Principal component analysis performed on children from group S and group N. Cumulative percentage variance explained By Axes: I – 27.30 %; I + II – 47.34%. Groups: S – Children who stutter; N- nonstuttering children. Variables: CCTT1 Time, CCTT1 failures, CCTT1 Warnings, CCTT2 Times, CCTT2 Warnings, CCTT2 number of sequencing errors, CCTT2 color sequencing errors, Anger/Frustration, Impulsivity, Sadness, Perceptual Sensitivity, Attention/Focusing, Soothability/Falling Reactivity, and age.

4. Discussion

This study investigated temperament dimensions, EF skills, and anxiety levels in children who stutter and their nonstuttering peers. The main results are consistent with the hypothesis that some children who stutter may differ in temperament and EF factors when compared to children do not stutter. Specifically, in these group comparisons, children who stutter were found to be more reactive and sensitive than their nonstuttering peers. However, the findings were different across the two age groups that were analyzed. The differences in temperament level were noted in the group of older children only, while differences in EF were noted in the group of younger children only. Furthermore, results did not support the idea that children who stutter exhibit higher rates of anxiety than children who do not stutter, regardless of age group. Correlation analyses highlighted the dynamic nature of stuttering and suggested

a link between endogenous abilities and external factors (Sudikoff et al., 2015; Wolfe and Bell, 2004).

4.1 Temperament

Results on the temperament scale are consistent with previous studies that have suggested difficulties in children who stutter compared to nonstuttering peers in attention span (Anderson et al., 2003; Costelloe et al., 2015; Eggers et al., 2010; Embrechts et al., 2000; Hollister, 2015) and a tendency toward impulsivity (Schwenk et al., 2007; Eggers et al., 2013). Attention and impulsiveness suggested a link to emotion regulation (Rothbart et al., 2001), because negative levels suggest emotional instability (Derryberry and Rothbart, 1988; Eisenberg et al., 1993). We also found differences in Anger/Frustration, Sadness, and Soothability/Falling reactivity temperament dimensions. This supports studies that indicate a more sensitive temperament in children who stutter. This could mean that school-age children who stutter may have more difficulty regulating their emotions. Furthermore, sadness could be connected to a more negative mood for children who stutter (Howell et al., 2004). A reactive temperament in children who stutter was also found in studies with preschoolers (Johnson et al., 2010; Ntourou et al., 2013) and school age children (Fowlie and Cooper, 1978). Higher scores in Anger/Frustration and lower scores in Soothability/Falling reactivity could indicate that older children who stutter (ages 10-12 years) can have more difficulty in recovering from peak distress, excitement, or general arousal (i.e., they may have a harder time settling down after an exciting activity) (e.g. Karrass et al., 2006).

4.2 Executive Functioning

Younger children who stutter required longer execution times and had a higher number of warnings and failures, number sequencing errors, and color sequencing errors compared age-matched peers who do not stutter. This suggests that children who stutter in the first years of schooling might have a lower attention span than their peers (Anderson et al., 2003). They might also need more time to adapt to a task and to start performing (Eggers et al., 2013; Manning and Beck, 2013) or have a greater concern about errors (Eichorn et al., 2017). A higher number of failures (times when a child almost makes a mistake) may be related to the tendency for impulsivity or difficulties with inhibitory control, as has been previously suggested by some authors

(Eggers et al., 2013; Schwenk et al., 2007; Ofoe et al., 2018). This was especially true for the task requiring the alternation of colors in the sequence of numbers.

4.3 Anxiety

No significant differences were detected between children who stutter and children who do not stutter in anxiety levels for either age group. According to previous studies, anxiety tends to increase as children grow older, especially between 8 to 12 years old (Blood and Blood, 2007; Messenger et al., 2015). These results are in agreement with prior researchers who reported no elevated anxiety in children who stutter (Mulcahy et al., 2008; Ortega and Ambrose, 2011; Smith et al., 2017). It could be that the participants in this study as a group showed no differences in anxiety because 22% were in speech therapy and another 22% had previously received treatment. Prior research has shown that people who are in or who have completed treatment often show comparable anxiety levels to their nonstuttering peers (Davis et al., 2002). Thus, balanced results between groups could be a consequence of the treatment itself. Other explanations may be due to methodological limitations, such as the lack of specificity of the measure to identify anxiety in the targeted population. As we saw above, anxiety in stuttering may be related to very specific situations, so, the use of a *trait* anxiety measure could have influenced the results. Speech tasks can trigger anxiety, so future research may benefit from using speech tasks rather than questionnaires (Gawda and Szepietowska, 2016; Manning and Beck, 2013). Finally, in self-report measures, children may try to give their answers a better view of themselves, trying to hide some perceived weaknesses and thereby under-reporting anxiety (Messenger et al., 2015).

4.4. Temperament, Executive Functioning and Anxiety interaction

Looking closely at the differences between groups, it was possible to observe different results in the older participants through the parent-perception scale and the younger participants through the performance of the EF task. It is hypothesized that Attention/Focusing, Perceptual Sensitivity, and Impulsivity issues may be subtle and unnoticed by the parents of the youngest children. Such differences may only be identifiable using sophisticated assessments such as the CCTT. In fact, some researchers agree that it is possible to find different results from behavioral measures (e.g. in novel events) and from parent reports of daily observations (Karrass et al., 2006). Moreover,

parent perspectives may not reflect children's true abilities (Bernstein Ratner and Silverman, 2000), because their responses may be influenced by the emotional link that exists with children (Seifer et al., 2004). Parents may also find it easier to identify temperament characteristics as children grow older, leading to more detailed or accurate assessment of children's temperament in the older age group. The results should be interpreted with caution since the sample was not matched by gender, with sex differences being related to the fact that more females were found in the schools where the sample collection, of children who stutter, was carried out. Finally, many tasks with different sensory modalities can also influence the results (Ofoe et al., 2018). In the present study, EF was assessed using a visual search task, but for the temperament results, parents may be basing their responses on situations that are dependent on other stimuli.

Because temperament characteristics can change over time (Rothbart et al., 2000), the different pattern between two age groups in temperament dimensions could also be related to the experience of negative emotional reactions and difficulties in functional communication abilities over time (Yaruss and Quesal, 2004; Yaruss, 2010). Current results from questionnaires may indicate that parents' responses are affected by experiences rather than an inherent tendency. As older children become more aware of their stuttering, by experiencing it in different situations, they may experience greater impact of stuttering in their lives. This might exacerbate or emphasize certain characteristics to the parents' view. When correlating the various components of temperament, EF, and anxiety, it was found that, difficulties in Attention/Focusing and Soothability/Falling reactivity were correlated with a tendency towards greater sadness and Anger / Frustration. Results are in agreement with previous literature (Sudikoff et al., 2015; Wolfe and Bell, 2004) which suggests an association between the coordination and integration of mental processes in successful task performance with self-regulation of emotional states (Sudikoff et al., 2015).

4.5 Temperament, Executive Functioning and Anxiety interaction and the DD-S model

Findings from the current study support the predictions from the DD-S Model (Walden et al., 2012), which state that cognitive and emotional regulation can be activated by exogenous contexts. According to the model, the cause of stuttering

moments is dynamic and not just related to external factors; it also relates to how children cope with exogenous factors through endogenous abilities (Walden et al., 2012). Further research on this dynamic relationship may be a starting point for better understanding the development of stuttering and the production of individual instances of disfluency. The present study helps to further specify the predictions of the DD-S model by the potential contribution of temperament and EF as intrinsic sensitivities, which can be triggered and boosted by external agents to influence the emergence of disfluencies.

4.6 Future Directions

Because endogenous capacities, such as temperament and EF, can change over time, and because exogenous factors, such as demands of the environment, may be different for each person, future research should examine the interactions between temperament and the development of EF both individually and over time. Similar studies that involve the analysis of several variables simultaneously may help to better explain the onset of anxiety in older children or other aspects of how stuttering - and reactions to stuttering - develop over time.

In future research, the use of multiple instruments would strengthen both the reliability and validity of these findings. For example, experimental methods that complement self-perception scales might allow the evaluation and analysis of child behavior in different situations. It would also be worthwhile to add inhibitory control and working memory tasks to better understand EF. These are the concepts that are encompassed in EF and have been examined independently in other studies (Eggers et al., 2013; Ntourou et al., 2017; Oyoun et al., 2010; Wolfe and Bell, 2004). The DD-S model predicts that emotional reactivity and emotion regulation influence the frequency and severity of stuttering in preschool-age children, so it would be appropriate for future research to examine these factors simultaneously.

Future studies should also employ a more balanced sample collection, with a more tight matching of groups in variables such as sex, age and other relevant factors. Although this study involved a reasonable sample size, the participants were in different stages of treatment, and it is possible that participants' treatment histories might have affected the results. Similarly, the presence of some differences in sex ratio and age

between sub-groups of children who stutter and children who so not stutter suggest that these preliminary results should be interpreted with caution.

5. Conclusion

Results highlight the potential role of emotional processes, temperament, and EF in the development of stuttering. Examining the cognitive and emotional skills of children who stutter across age groups can add further knowledge about stuttering. Ultimately, such knowledge may lead to refinements in clinical and educational practices. A principal outcome of this study is the finding that endogenous abilities in children who stutter may be different according to their age. Older participants were found to be more prone to difficulties in temperament dimensions, while younger participants exhibited predispositions for difficulties related to EF. This suggests that differences between children who stutter and children who do not stutter may be mediated by age and development. These results are in agreement with a dynamic view of the development of stuttering influenced by internal and external factors.

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CHAPTER 3: Portuguese school-age children's experience of stuttering[3]

[3]The following study is under review and awaiting for acceptance: Rocha, M.S., Yaruss J.S. & Rato J.R. (under review). Portuguese school-age children's experience of stuttering. *Speech, Language and Hearing.*

Abstract

Stuttering is a multifaceted disorder that can affect children's psychological state, academic performance, social relationships, and quality of life. Therefore, it is crucial to explore the impact of stuttering in children, based to their own perspectives and experiences. The purposes of this study were to: (a) investigate the impact of stuttering in Portuguese school-age children and (b) evaluate the reliability and validity of the European Portuguese translation of Overall Assessment of the Speaker's Experience of Stuttering (OASES-S-PT). Participants were 50 Portuguese children who stutter aged 7 to 12 years (M=9.10, SD=1.7). Overall, participants exhibited a mild to moderate overall impact from stuttering. Results suggest that the OASES-S-PT is a suitable measure for assessing the impact of stuttering on Portuguese children. A comparison of adverse impact with OASES data in other countries highlighted the need to include different cultural perceptions in the research about stuttering experiences.

1. Introduction

Although widely studied, stuttering continues to be considered as a complex and multi-factorial disorder that is influenced by environmental, genetic, and constitutional factors (Smith & Weber, 2017; Yaruss & Quesal, 2004). Previous research has shown that stuttering is often associated with negative reactions, difficulties with daily communication, and adverse impact on overall quality of life (e.g. Craig, Blumgart, & Tran, 2009; Klompas & Ross, 2004; Yaruss & Quesal, 2006; Yaruss, 2010).

Whereas it was long believed that such negative consequences primarily affected older individuals who stutter, more recent research has shown that children can also develop negative attitudes about stuttering (Ambrose & Yairi, 1994; Boey et al., 2009; Clark, Conture, Frankel, & Walden, 2012; Murphy, Yaruss & Quesal, 2007; Pukacova, 1973; Vanryckeghem, Brutten, & Hernandez, 2005; Yovetich, Leschied, & Flicht, 2000).For example, research shows that even very young children who stutter may perceive themselves as poorer speakers and have lower self-esteem (e.g. Boey et al., 2009, Langevin, 2009, Pukacova, 1973; Yovetich et al, 2000;Vanryckeghem, Hylebos, Brutten, & Peleman, 2001). The way in which this impact arises is not well-documented, particularly for children (e.g. Davis, Howell, & Cooke, 2002; Murphy et al., 2007; Vanryckeghem et al., 2001; Blood & Blood, 2004; Yaruss & Quesal, 2016). Nevertheless, there is consensus in the literature that children may negatively react to

their speaking difficulties, experience challenges with social interaction, and be subject to bullying and other negative experiences that can adversely affect their quality of life. Such experiences may negatively affect children's full participation in educational, future vocational and recreational opportunities (Blood & Blood, 2004; Davis et al., 2002; Hugh-Jones & Smith, 1999; Murphy & Quesal, 2002; Murphy et al., 2007).

Feelings and thoughts about speech, and about children's lives as a whole, play an important role in the diagnosis, intervention, and prognosis of stuttering. Consideration of these factors helps in assessing the impact of stuttering and in determining the need for treatment (e.g. Conture, 2001; Gregory, 2003; Reardon-Reeves & Yaruss, 2013; Yairi & Seery, 2010; Zebrowski & Kelly, 2002). Therefore, in addition to assessment of speech behaviors in children who stutter, it is crucial to also assess the impact of stuttering on quality of life (Beilby, 2014; Erickson & Block, 2013;Yaruss & Quesal, 2004a, 2006). The World Health Organization (WHO, 2001) relates quality of life to individual perceptions about several aspects of a person's life, including culture and value systems. Children's quality of life may be influenced by their goals, expectations, standards, and concerns within the context of their own society. These, in turn, influence and affect a children's physical health, psychological state, level of independence, social relationships, and personal beliefs, among other aspects of life. Despite the importance that has been given to research on quality of life in pediatric disorders (Solans et al., 2008), little is known about the impact of speech and language disorders, specifically in stuttering (Feeney, Desha, Ziviani, & Nicholson, 2011)

Grims (1978) and Guitar & Grims (1979) were among the first to propose a scale that directly assessed the attitude of school-age children about their speech. The A-19 scale involves 19 speech-specific statements about children's experiences in situations that typically involve speech, where children are asked to answer "yes" or "no" to the scale questions. Although the tool has long been available, validity data is still being collected (Guttormsen et al., 2015). Other instruments have been also developed over the years to assess various aspects of the stuttering disorder. For example, the *Communication Attitude Test* (CAT; Brutten & Vanryckeghem, 2006) is a self-report test that encompasses 35 true/false statements about speech-associated attitudes of school-age children who stutter. Analysis of comparative and inter-item reliability shows that the CAT is a valid instrument with strong reliability that can be used by both researchers and clinicians to evaluate how children think and feel about their stuttering (Brutten & Vanryckeghem, 2006). Both the CAT and A-19 scales

address communication attitudes; however the CAT is more extensive, as it was designed to account for attitudes differences between children who stutter and their non-stuttering peers (Bernardini, Vanryckeghem, Brutten, Cocco, & Zmarich, 2009; Guttormsen et al., 2015).

A widely used instrument that intends to provide a comprehensive view of the stuttering disorder in school age children is the *Overall Assessment of the Speaker's Experience of Stuttering – School-Age (Ages 7-12)* (OASES-S; Yaruss & Quesal, 2016). The instrument is based on the World Health Organization's International Classification of Functioning, Disability, and Health (ICF; World Health Organization [WHO], 2001), as adapted to stuttering by Yaruss (1998) and Yaruss and Quesal (2004). The ICF is comprised of several components, including body function and structure, activities and participation, and personal and environmental context. 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 (Yaruss & Quesal, 2016), as well as in translated versions around the world (Beilby, Byrnes, & Yaruss, 2012; Blumgart, Tran, Yaruss, & Craig, 2012; Bodil, Sønsterud, & Kirmess, 2018; Chun, Mendes, Yaruss, & Quesal, 2010; Euler, Anders, & Merkel, 2016; Freud, Kichin-Brin, Ezrati-Vinacour, Roziner, & Amir, 2017; Koedoot, Bouwmans, Franken, Stolk, 2011; Lankman, Yaruss, & Franken, 2015; Osborn, Yaruss, Quesal, Schiefer, & Chiari, 2012; Sakai, Chu, Mori, & Yaruss, 2017; Yadegari et al., 2018). Results suggest that the OASES instrument may be suitable for both clinical and research use in providing quantitative and qualitative information about the impact of stuttering on the lives of children, adolescents, and adults who stutter.

The original normative data for OASES-S involved data from 75 school age American children, with an average age of 10 years; almost half of the children (48%) reported a moderate impact of stuttering in their lives (Yaruss & Quesal, 2004a). Normative data for OASES-S has been collected in the Netherlands (Lankman et al., 2015) and Australia (Beilby et al., 2012). In the Netherlands (n=101), the majority of children (60.2%) exhibited mild-to moderate impact of stuttering in the overall scores. In Australian (n=50), the majority of children exhibited a moderate-to-severe impact (Beilby et al., 2012). There is also a study with a small sample in Brazil (n=7), in which approximately half of the children studied were classified as "moderate" in the overall

impact (Chun et al., 2010). All children in this studies were engaged in treatment and were in different levels of stuttering severity (Beilby et al.,;Lankman et al., 2015; 2012Yaruss & Quesal, 2004a).While in Australia children were waiting for treatment, in EUA and Netherlands children were in different stages of treatment. There is no information about children recruitment in Brazil.

As children grow, stuttering may continue to be a burden in several aspects of life and interfere with the individual's ability to participate in daily activities (Craig et al, 2009, Yaruss & Quesal, 2004b). Several studies have demonstrated the impact of stuttering on quality of life in adolescents and adults (e.g. Klein & Hood, 2004; Klompas & Ross, 2004; Koedoot, Bouwmans, Franken, & Stolk, 2011). Adolescents and adults who stutters may continue to experience negative affective, behavioral, and cognitive reactions from themselves and from the people in their environment (Yaruss & Quesal, 2004a). These reactions, which may include embarrassment, shame, fear, low self-esteem and self-confidence, among others (e.g. Klein & Hood, 2004; Klompas & Ross, 2004).

Stuttering affects people all around the world; however perceptions about the disorder and the way of seeing may differ by culture. Studies have revealed that some stereotypes, beliefs, and attitudes are consistent across countries, while other beliefs are region- or culture-specific, especially those related to religious causes (e.g. St. Louis et al., 2016). Using the Public Opinion Survey of Human Attributes-Stuttering (POSHA-S), St Louis et al. (2016) and Valente et al. (2017) found notable differences between countries and cultures across Europe. Specifically, they reported more negative attitudes in Italy, and more positive attitudes in Norway and Sweden, compared with the averages in the POSHA-S database. In Germany, Poland, and Portugal, public stuttering attitudes are closest to the median values (St. Louis et al., 2016). Such cross-cultural comparisons provide meaningful information about the perceptions of stuttering by listeners; similar information about the experiences of stuttering by speaker is also valuable. To the best of our knowledge, however, there are no European Portuguese instruments for assessing either the overt or covert aspects of stuttering. Such measures should address linguistic and cultural differences, as prior research has shown differences between countries.

For studies involving the OASES, for example, modifications were made to the original English versions for studies in the Netherlands and Brazil account for specific vocabulary that is commonly used into the language of each country (Chun et al., 2010;

Lankmanet al, 2015). Even when countries share the same language, there may be notable differences between them in terms of cultural identity and language evolution. This is also the case for the Portuguese language spoken in Brazil and Portugal (Silva, 2018).

In order to understand how Portuguese children experience stuttering and its impacton their lives, we conducted in the present study the Portuguese translation of the *Overall Assessment of the Speaker's Experience of Stuttering* for School-Age children (OASES-S-PT). In addition, we also take into account potential cross-cultural factors to investigate the reliability and construct validity of this instrument, considering the data of the versions used in other countries.

2. Methods

2.1 Participants

Participants of this study were 50 Portuguese school-age children (36 male and 14 female), ages 7 to 12 years old, with mean age of 9 years 10 months old (SD=1.7 years). The sex ratio of participants who stutter was 2.6 boys to each girl. This figure is generally consistent with previously reported values (e.g. Craig, Hancock, Tran, Craig, & Peters, 2002; Yairi & Ambrose, 2005). The sample includes only children without any neurological impairment (other than stuttering), psychiatric disturbance, or history of head injury, learning disorder, or seizure. The sample was chosen by convenience, in Portugal, and collected from speech-language therapist cases and through referral of teachers and other professionals.

The sample consists of children who were in speech therapy at the time of the study (n=25), children who had previous speech therapy (n=11), and children who were waiting for therapy or just initiating speech therapy (n=14). The children who were in therapy at the time of data collection had been in treatment between 1 month to 96 months (M= 9.30 mos.; SD = 19.38 mos.). Children who had previous therapy had received between 3 months and the 48 months of treatment (M=13.28 mos.; SD = 12.99 mos.).

2.2 Materials

The Stuttering Severity Instrument – 4th Edition (SSI-4; Riley, 2009), with the Portuguese story, "*A história do rato Artur*" (Guimarães, 2002), was used to confirm the diagnosis of stuttering. The parents provided information about child's stuttering and socio-demographic background via a checklist created for this study, gathering information about participant's gender, history of prior therapy, education level, and family history of stuttering.

Researchers developed a new translation and adaptation of the *Overall Assessment of the Speaker's Experience of Stuttering – School-Age* in European Portuguese (OASES-S-PT; *Avaliação Global da Experiência da Gaguez pela Pessoa – Versão Portuguesa*; Rocha et al., 2019). Like the original English OASES-School-age (OASES-S) instrument, the OASES-S-PT instrument assesses the adverse impact or negative consequences associated with stuttering. It is divided in 4 sections. Section I (General Information), contains 15 items pertaining to the speakers' perceived fluency and speech naturalness, knowledge about stuttering, and overall feelings about stuttering; Section II (Your Reactions to Stuttering), contains 20 items examining the speakers' affective, behavioral, and cognitive reactions to stuttering; Section III (Communication in Daily Situations) contains 15 items assessing the degree of difficulty speakers have when communicating in general situations, at school, in social situations, and at home; Section IV (Quality of Life) contains 10 items assessing how much stuttering interferes with the speakers' satisfaction with their ability to communicate, their ability to participate actively in life, and their overall sense of well-being.

2.3 Procedures

This study received full ethical approval by the Ethics Committee of Institute of Health Sciences of Universidade Católica Portuguesa, register number 34/2017, approval by one of the OASES-S authors, and permission from the publisher of the OASES (Stuttering Therapy Resources, Inc.). Prior to their participation in this study, parents provided informed consent for themselves and their children. Consent also

included permission to audio/video record the child. The right to withdraw from the study at any time was clarified.

The OASES-S was translated to European Portuguese (OASES-S-PT) using a forward/backward translation process, as follows: First, the English OASES-S was translated taking into account linguistic, cultural, and contextual issues. This was carried out by two Portuguese speakers who are fluent in English. After this procedure, a consensus version of the translated versions was made. A document containing the main differences between the two versions was prepared, and it was possible to verify that these differences were mainly semantic (synonymous words) or morphosyntactic (slight changes in the sentence constructions). The next step included a reverse translation carried out by one Portuguese speaker fluent in English (different evaluator from the original translators). Finally, another Portuguese speaker fluent in English (again, different from the others) compared the original version and the reverse-translated version, finding, once more, minor semantic and morphosyntactic differences. A further adaptation of the OASES-S-PT questions was completed and the two versions were reviewed by a fluency specialist Speech Language Therapist.

A pilot study was conducted using the questionnaire on a small sample of 5 children, who were attending therapy at that time (mean age = 8 years, 0 months; SD = 1.41 years) and were not included on the sample of the 50 children. The results revealed that 7-year-old children had difficulties in reading OASES-S-PT alone. Therefore, in the application of the instrument to the sample, the survey questions were read in full by the researcher in charge of the assessment. For older children, the questions were read aloud and explained whenever requested, following the original procedures described in the OASES manual (Yaruss & Quesal, 2016).

After the pilot study, we started the evaluation of the 50 children in the sample for this study. The researcher assessed the child with the OASES-S-PT, while, in a different room sociodemographic checklists were completed by parents. Children took about 15 minutes to complete OASES-S-PT questionnaire. All testing was conducted between December 2017 and June 2018.

3.3.1 Data analysis

Several statistical procedures were performed in order to analyze reliability and validity of OASES-S-PT. Data analysis was conducted with SPSS (Statistical Package

for the Social Sciences -Version 24 for windows; Armonk, NY: IBM Corp). Two forms of reliability were assessed: internal consistency reliability and test-retest reliability. To assess test-retest reliability, the OASES-S-PT was re-administered to 8 children who stutter (16% of the total sample of 50 participants), identified through professional and clinical contacts of the author, with an interval of 15-30 days from the initial administration. Participants did not receive any treatment in the intervening period. Paired sample t-tests were used to compare mean scores from the two applications. In order to support the internal consistency reliability and the construct validity of OASES-S-PT, Cronbach's alpha and OASES scores were related to one another and to other variables, using Pearson's correlation coefficient. The distribution of impact scores for Portuguese children ages 7-12 was then compared to the results from other countries, through two-sample t-tests in order to compare scores means.

3. Results

3.1 Characteristics of OASES-S-PT

Table 1 reports the mean, mode, standard deviation (SD), skewness, and kurtosis of the items in the four OASES-S-PT sections.

For section I (General information), there were no scores indicating less than mild to moderate impact (Min = 1.98). The overall mean score corresponds to a moderate to severe impact (M=3.03; SD=0.63). For section II (Reactions to stuttering), there were no scores below mild to moderate impact (Min=1.52) or higher than severe impact (Max=2.87). The overall mean score corresponds to a mild to moderate impact (M=2.10; SD=0.40). In section III (Communication in daily situations), the minimum score reflected a mild impact (Min=1.12), and no scores exceeded a moderate impact (Max =2.50). For section III, the overall mean score corresponds to a mild to moderate impact (M=1.68; SD =0.43). Finally, Section IV (Quality of Life), the minimum, maximum and mean scores all correspond to the same impact scores as section III (Min = 1.20; Max = 2.64; M=1.66; SD =0.43). Overall, scores ranged from 1.66 and 3.03, with lower mean scores for section IV and higher mean scores for section I.

Examination of the distributions of the scores revealed asymmetry. Skewness ranged between -0.22 and 1.68, with a positive skewness for all sections except section I. Kurtosis ranged between -0.60 and 2.56. Although the distributions were not normal, the participants' responses exhibited acceptable variability.

Table 1: Descriptive statistics for OASES-S-PT sections items.

Item Number	Mean	Mode	SD	Skewness	Kurtosis
Section I: General Information					
1	2.82	3	0.77	0.05	0.79
2	2.56	3	1.18	0.39	-0.46
3	3.42	6	2.00	0.25	-1.56
4	1.98	1	1.10	0.71	-0.48
5	3.22	3	1.20	-0.00	-0.48
6	3.34	5	1.47	-3.40	-1.16
7	3.22	3	1.41	-0.09	-1.15
8	3.42	3	1.39	-0.33	-0.95
9	4.50	5	0.95	-1.92	3.16
10	2.44	3	0.97	0.11	-2.96
11	2.22	2	1.01	0.87	0.71
12	3.16	2	1.99	0.56	-1.38
13	2.88	3	1.11	-0.28	-0.78
14	3.68	4	1.08	-0.64	-0.07
15	2.64	2	1.08	0.38	-0.33
Mean	3.03	3.20	1.25	-0.22	-0.47
SD	0.63	1.32	0.36	1.10	1.36
Min	1.98	1.00	0.77	-3.40	-2.96
Max	4.50	6.00	2.00	0.87	3.16
Section II: Your Reactions to Stuttering					
16	2.66	3	0.96	-0.40	-0.70
17	2.34	1	1.19	0.36	-0.84
18	2.62	3	1.21	0.14	-0.90
19	2.28	1	1.20	0.32	-1.17
20	1.52	1	0.81	1.35	0.71
21	2.40	1	1.29	0.50	-0.81
22	1.60	1	1.05	1.99	3.45
23	2.02	1	1.13	0.75	-0.53
24	1.94	1	1.17	1.00	0.08
25	1.68	1	1.02	1.41	1.31
26	1.90	1	1.07	0.82	-0.26
27	2.06	1	1.10	0.56	-0.67
28	1.72	1	0.83	0.80	-0.43
29	1.90	2	0.84	0.84	0.45
30	1.86	1	1.05	1.17	0.67
31	1.78	1	0.89	1.19	0.96
32	2.78	1	1.49	0.09	-1.53
33	1.84	1	0.98	1.29	1.50
34	2.86	1	1.43	-0.01	-1.37
35	2.18	1	1.27	0.82	-4.85
Mean	2.10	1.25	1.10	0.75	-0.60
SD	0.40	0.64	0.19	0.57	2.45
Min	1.52	1.00	0.81	-0.41	-8.09
Max	2.87	3.00	1.49	1.99	3.45
Section III: Communication in Daily Situations					
36	1.50	1	0.54	0.40	-1.05
37	1.72	1	0.78	0.81	-0.00
38	1.60	1	0.70	1.12	1.47
39	2.44	1	1.25	0.27	-1.14
40	1.28	1	0.64	2.60	7.02
41	2.10	1	1.13	0.77	-0.44
42	1.80	1	0.90	0.76	-0.51
43	1.84	1	1.13	1.03	-0.44

44	1.40	1	0.70	1.86	3.31
45	1.26	1	0.57	2.83	10.32
46	1.92	2	0.94	1.07	1.21
47	1.58	1	0.79	1.70	3.29
48	1.12	1	0.33	2.41	3.97
49	2.50	1	2.23	0.96	-1.07
50	1.18	1	0.44	2.45	5.73
Mean	1.68	1.07	0.87	1.40	2.11
SD	0.43	0.26	0.46	0.84	3.46
Min	1.12	1.00	0.33	0.27	-1.14
Max	2.50	2.00	2.23	2.83	10.32
Section IV: Quality of Life					
51	2.08	1	1.06	0.93	0.64
52	1.72	1	0.99	1.39	1.51
53	2.64	1	2.26	0.81	-1.30
54	1.48	1	0.86	2.05	4.63
55	1.42	1	0.84	2.34	6.16
56	1.20	1	0.50	2.53	5.85
57	1.60	1	0.90	1.42	1.07
58	1.22	1	0.51	2.31	4.77
59	1.60	1	0.95	1.35	0.55
60	1.60	1	1.06	1.63	1.68
Mean	1.66	1	0.99	1.68	2.56
SD	0.43	0	0.49	0.60	2.58
Min	1.20	1	0.50	0.81	-1.30
Max	2.64	1	2.26	2.53	6.16

Table 2 reports the internal consistency reliability and standard error of measurement (SEM) data for OASES-S-PT. Analyses revealed a high degree of reliability (Cronbach's alpha ranging from 0.84 to 0.96) and low SEM (ranging from 0.14 to 0.27).

Table 2: Internal Consistency Reliabilities and SEMs of Impact Scores for the OASES-S-PT (N=50).

Section	Mean	SD	Reliability (α)	SEM
I: General Information	3.03	0.63	0.84	0.26
II: Your Reactions to Stuttering	2.10	0.40	0.93	0.12
III: Communication in Daily Situations	1.68	0.43	0.88	0.15
IV: Quality of life	1.66	0.41	0.88	0.14
Overall Score	2.16	0.71	0.96	0.15

Test-retest reliability was calculated based on: a) the correlation between scores for the two administrations, b) point-to-point agreement and, c) the average absolute difference between individual item scores and between impact scores. Table 3 presents

the data for the 8 children who participated in the test-retest analysis. Point-to-point agreement analyses, which examined the degree to which individuals provided the same responses to individual items form one administration to the next, revealed that 90.83 % of the item responses were identical and 98.75% of the responses were within one point of each other.

Table 3: Test-retest Reliabilities of Impact Scores for the OASES-S-PT

	First Testing		Second Testing			
Section	Mean	SD	Mean	SD	*t*	*p*
I: General Information	2.69	0.79	2.87	0.59	-1.94	0.09
II: Your Reactions to Stuttering	2.25	0.80	2.16	0.68	1.20	0.27
III: Comm. in Daily Situations	1.41	0.35	1.77	0.71	-1.49	0.18
IV: Quality of life	1.63	0.70	1.58	0.66	1.01	0.35
Overall Score	2.06	0.57	2.05	0.50	0.27	0.78

Note. N=8; interval = 15 to 30 days

The average absolute differences in item and impact scores between the two administrations are reported in Table 4. The average absolute differences between item scores and impact scores were generally very small (ranging from 0.08 to 0.16 and 0.08 to 0.38, respectively). These results are not statistically significant and they don't have any impact on clinical practice.

Table 4: Mean Absolute Test- Retest Differences in Item and impact Scores for the OASES-S-PT

Section	Item Score	Impact Score
I: General Information	0.16	0.23
II: Your Reactions to Stuttering	0.09	0.16
III: Communication in Daily Situations	0.08	0.38
IV: Quality of life	0.09	0.11
Overall Score	0.10	0.08

Construct validity data are presented in table 5. To verify that each section of the test examined a different construct, Pearson's correlations were calculated between impact scores on the sections. The results showed a moderate correlation (ranging from 0.44 to 0.59) between sections and an expected higher correlation (ranging from 0.74 to 0.90) between sections and the overall score.

Additional evidence of the validity of test scores, such as concurrent validity and criterion validity, is typically provided through correlations with other tests that are thought to be good measures of the constructs of interest. To date, however, there were no other Portuguese published questionnaires to assess the speaker's experiences of stuttering in school age children to conduct this analysis.

Table 5: Correlations among section Impact Scores for the OASES-S-PT ($p < 0.01$)

Section	I	II	III	IV
I: General Information				
II: Your Reactions to Stuttering	0.59			
III: Communication in Daily Situations	0.63	0.52		
IV: Quality of life	0.44	0.58	0.56	
Overall Score	0.80	0.90	0.77	0.74

3.2 OASES-S scores for Portuguese children who stutter

Figure 1 shows the distribution of scores for all four sections and for the Overall Impact ratings.

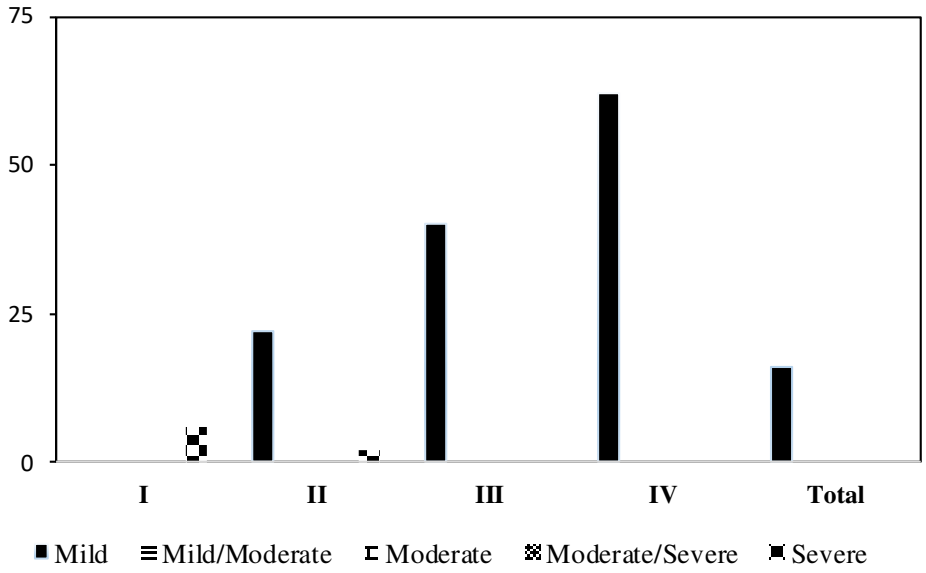

Figure 1. Distribution of impact rating sections.

The results show that the majority of participants (44%) had a moderate overall impact rating of stuttering. A notable percentage (36%) of participants had a mild to moderate impact, and a small percentage of participants had a mild (16%) or moderate-to-severe impact (4%). In our study we did not find participants with severe impact.

Examination of the four sections of the OASES-S-PT revealed that participants had higher negative impacts in section I (42% of children had a moderate severe impact) and lower negative impact in section IV (62% of children had a mild impact). In section I, there were no participants with a mild impact, and in section III and IV there were no participants with a severe impact.

3.3 OASES-S scores from around the world

In order to perform a cross-cultural comparison, these European Portuguese data were compared with the data obtained in the other countries, as presented in published works (Beilby et al., 2012; Lankman et al., 2015; Yaruss & Quesal, 2016).

Table 6 reports results from a comparison of the present data with the original standardization data in the USA and revealed statistically significant differences in section I ($t_{(77.013)} = 2.98$, $p = .004$) and III ($t_{(113.463)} = -3.422$, $p = .001$), with Portuguese children scoring higher (indicating greater impact on issues related the general perception of stuttering and in terms of functional communication difficulties). No statistically significant between-group differences ($p > .05$) were found in sections II and IV.

A comparison of the present data with those from the Australian study revealed statistically significant differences in three of the four sections, with lower scores observed for Portuguese children in sections II ($t_{(77.515)} = -8.62$, $p < .001$), III ($t_{(98)} = 18.12$, $p < .001$) and IV ($t_{(91.639)} = -12.83$, $p < 0.001$).

In the comparison between two European countries, statistically significant differences were found in section III, with lower scores for Portuguese ($t_{(119.911)} = -2.87$, $p = .005$) compared with Dutch children.

Table 6: Distribution of means (M), standard deviations (SD) and t-test comparison of OASES-S between Portugal and USA, Western Australia and Netherlands.

Countries (N)	Section I		Section II		Section III		Section IV	
	M (SD)	*t*	M (SD)	*t*	M (SD)	*t*	M (SD)	*t*
USA (75)	2.57 (0.40)	2.98**	2.38 (1.06)	-1.39	2.08 (1.06)	-3.42**	1.72 (0.93)	-1.42
W. Australia (50)	3.08 (0.44)	-1.78	3.24 (0.44)	-8.62**	3.30 (0.43)	18,12**	2.97 (0.48)	-12.83**
Netherlands(101)	2.75 (0.40)	1.29	2.04 (0.57)	0.90	1.87 (0.64)	-2.87**	1.57 (0.55)	-0.35
Portugal (50)	3.03 (0.63)	-	2.10 (0.40)	-	1.68 (0.43)	-	1.66 (0.41)	-

**p < 0.01

4. Discussion

This study revealed that the European Portuguese translation of the OASES-S is a reliable and valid instrument for evaluating the impact of stuttering on the lives of Portuguese children. Specifically, data indicated a high point-to-point and average score agreement in test-retest reliability, strong correlations within sections, and a low *SEM* for all measures. These results support its use for repeated administration in clinical

settings and treatment outcomes studies. Moreover, the correlations within sections were higher than the correlations between sections with good internal consistency, thereby supporting the notion that the sections measure different constructs.

With the OASES-S-PT, we found that Portuguese children who stutter, engaged in treatment, experienced a moderate overall adverse impact because of their stuttering. This was particularly evident in Section I, which revealed that children have a low overall knowledge or self-awareness of stuttering and may have negative feelings about speech and stuttering. Considering that most of the participants in this study were either currently in treatment or had previously received treatment, one would have expected these participants to be more knowledgeable about stuttering. In addition, participant's results suggest difficulties in accepting stuttering and negative attitudes toward speaking and stuttering. However, in section III and IV there were no participants with a severe impact, which once again, can be related to previous treatment.

These results are in agreement with the fact that there are few studies and little investment in stuttering research and treatment in Portugal. This might help to explain the relative lack of knowledge about the subject in the general population. Still, results are consistent with findings from previous studies, especially those examining children in Australia (Beilby et al., 2012) and the Netherlands (Lankman et al., 2015). In the USA, the impact reflected in Section I was notably lower (Yaruss & Quesal, 2004a). Our results on Section I may indicate the need to refine therapeutic programs in Portugal so they include more information and education about stuttering. Treatment in Portugal may also benefit from a greater focus on reducing unfavorable impressions about speaking ability as opposed to focusing primarily on speech or stuttering modification (Berquez & Kelman, 2018, Murphy et al., 2007).

The results from Section II (Reactions to Stuttering) confirm that Portuguese school-age children who stutter can experience negative emotional reactions, including embarrassment, frustration, or anxiety associated with stuttering. They can also exhibit signs of physical tension and have difficulties accepting feelings that arise from stuttering. These findings were similar to those from the USA and the Netherlands, though they differed from the findings in Australia. Children in this country Autralia showed an even higher impact than children in the other samples. However, it sould be noticed that children in Autralia were all in a waiting list for treatment. Again, these results may indicate a need to incorporate more work on desensitization to stuttering in order to reduce negative reactions to disfluent speech and physical tension. However,

the mismatch observed in the first two sections indicate that the focus on knowledge increment should be more strengthened in the therapeutic process.

In Section III (Communication in Daily Situations), participants reported being able to communicate freely in several scenarios, like as at school, at home, and in social settings; however, it was clear that some situations were harder than others. For example, speaking in small groups was rated as easier than speaking in large groups. This is in agreement with other studies (Blood & Blood, 2004; Davis et al., 2002; Hugh-Jones & Smith, 1999; Murphy & Quesal, 2002; Murphy et al., 2007). Comparing to other countries, Portuguese children showed a minor impact of stuttering in this section. This may be associated with the fact that Portuguese therapeutic programs (like others) often include avoidance reduction in their treatment goals, as well as strategies to improve fluency in difficult situations.

Section IV (Quality of Life) revealed that Portuguese children experience minimal impact on their overall quality of life associated with stuttering. These data are consistent with data from the USA and the Netherlands (Lankman et al., 2015; Yaruss & Quesal, 2016). Children in Australian children experience greater impact on their quality of life (Beilby et al., 2012).

The results of participants in this study suggest that there may be benefits to increasing the emphasis on desensitization and acceptance in treatment; with such changes, the impact of stuttering may decrease further.

4.1 Limitations and future directions

Potential limitations of the current work are similar to those seen in other studies that use self-report measures, including the possibility of response bias. However, self-report methods are one of few methodologies available for collecting information about a speaker's attitudes and emotions regarding their own communication. Subsequent research might consider qualitative methods to verify and validate the self-report responses of children who stutter. Although this study involved a reasonable sample size, the sampling methods may have influenced the outcomes .The participants were in different stages of treatment, and it is possible that participants' treatment histories might have affected their answers. It would be appropriate to replicate this study using a larger sample of school-age children divided into balanced groups based on prior treatment to better understand the potential impact of treatment.

Finally, results from the comparison between countries should be interpreted with caution. Despite the sampling methods are quite similar, some diferences can be found and should be considered in future studies.

Prior to the present study, there were no Portuguese instruments to assess and analyses stuttering and its impact on children's lives. Results showing that the OASES-S-P is a reliable and valid measure for assessing the impact of stuttering in children's lives provides a starting point for additional studies aimed at improving our understanding of the experiences of children in Portugal who stutter and for developing customized interventions that address the unique needs of individual children who stutter based on the ways in which stuttering affects their lives.

5. Conclusions

The OASES-S-PT appears to be a reliable and valid instrument for providing a comprehensive and holistic assessment of the impact of stuttering in school-age Portuguese children who stutter. The present study shows that children who stutter can experience negative impact in their lives due to their stuttering and to the ways in which they perceive their speaking difficulties. Findings revealed that Portuguese children experience a moderate impact of stuttering in their lives. Even if it is not possible to make a direct and valid comparison between countries, the differences found reinforce the value in considering different cultural perceptions.

Further studies along these lines will contributes to a better understanding of the complexity of stuttering and support the development of individualized intervention programs. With the study of OASES in several countries, treatment programs can take a comprehensive, biopsychosocial approach and be personalized, as appropriate, to each person and culture.

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CHAPTER 4: Stuttering impact: A shared perception for parents and children?[4]

[4] The following study is under review and awaiting for acceptance: Rocha, M.S., Yaruss J.S. & Rato, J.R. Stuttering impact: A shared perception for parents and children? *Folia Phoniatrica et Logopaedica.*

Abstract

Previous research has provided information about how school-aged children perceive their own stuttering; however, less is known about how stuttering is perceived by their parents. The ways that parents view their children's stuttering could influence how the children themselves react to it. This study proceeds to assess how parents' perceptions of the impact of stuttering relate to the perceptions of children.

Participants were 50 children who stutter aged 7 to 12 years (M=9.10; SD=1.7) and their parents, recruited from different cities in Portugal. The European Portuguese version of Overall Assessment of the Speaker's Experience of Stuttering (OASES-S-PT) was administered to the children, and an adapted version of the tool was administered to their parents.

Both parents and children showed generally similar overall impact ratings, typically falling in the mild and moderate ranges. Differences were observed in families with a history of stuttering: for those families, a comparison of parents' and children's scores revealed, in some domains, that parents perceived the impact of stuttering to be greater than the children did, specially related with children's reactions to stuttering and their quality of life.

Knowledge about how parents perceive the impact of stuttering on their children is important because families can play a key role in helping children cope with stuttering. These findings highlight the benefits of resorting to an individualized treatment approach for each child that focuses on their perceptions, as well as on those of the parents, in order to work on negative attitudes toward children's stuttering.

1. Introduction

Stuttering is typically defined as a disruption of communication, characterized by involuntary interruptions in speech, such as repetitions, prolongations, and blocks [1]. The core features of stuttering that are included in such definitions are those that are more easily perceived by the interlocutor; however, stuttering is a complex phenomenon that also involves emotional components, including stress, anxiety, and negative reactions to speaking [1–4]. Thus, stuttering may be seen as a multi-factorial disorder encompassing several aspects liable to influence the onset, manifestation, and chronicity of stuttering in each individual child [2,5,6].

In recent decades, professionals within this field have come to a deeper understanding of how stuttering might adversely affect children. One way to explain these broader effects of stuttering is through the International Classification of Functioning and Disability and Health (ICF), part of the World Health Organization (WHO) [7]. The ICF provides a comprehensive view of the disorder, describing the ways in which the stuttering impairment may limit children's communicative ability and limit their participation in daily activities [4, 9].

In an attempt to ensure a comprehensive assessment, professionals within this field have also focused on parents' contributions regarding the kind of therapy aimed at children who stutter [9, 10, 11]. In keeping with a multidimensional view of the disorder, it is relevant to combine the observations and experience of parents alongside the expertise of professionals, when considering the ways in which stuttering affects a child's life [9,11,12]. Parental input is also important due to the variability of stuttering, since they can give relevant information about children's speech fluency and communication in situations to which clinicians aren't otherwise able to access [13]. Furthermore, understanding family dynamics and parents' attitudes toward stuttering could improve the management of stuttering in children [4,14,15]. Family relationships have been found to affect the children who stutter in the ability to cope with stuttering, once families can provide support and positive guidance to their children. Therefore, it should be a priority for clinicians to understand how home environment influence the development of stuttering [15,16].

Over the years, several measures have been developed for assessing the stuttering disorder. Some of these measures are primarily focused on the recognisable features of stuttering, while others address feelings, attitudes, thoughts, and reactions. For example, the *Communication Attitude Test* (CAT) [17] and the *Overall Assessment of the Speaker's Experience of Stuttering – Ages 7-12* (OASES-S) [18] both measure the ways in which children react to stuttering. Because children may not have a full understanding of the ways in which stuttering might affect them, however, it is also important for clinicians to gather information from parents. This can be done through informal interviews or through formal scales, such as the *Palin Parent Rating Scales* [9], and observational rating scales, such as those included in the *Test of Childhood Stuttering* (TOCS) [9,19,20]. These measures may provide information about the child's behavioural and emotional reactions to stuttering beyond the clinic context [21–23].

The information provided by parents may be different from that provided by the child and such variance in ratings may be due to different perspectives about who has the experience [11, 25]. For example, research on various conditions, such as emotional-behavioural disorders, has shown that parents' and children's responses may be significantly different [25,26]. In the field of stuttering, there are authors who claim that despite the importance of parents, in the whole process of treatment, they not appear to be valid informants [24]. However, some studies in pediatric area, for example, behaviour and mental health just reveal slight differences [26–30]. Some authors [26] point out that parents' responses may be influenced by the desire to please the examiner or to respond as they think is socially acceptable. Others [26, 30] highlight the influence of parents' comparisons with their other children or a parents' lack of knowledge about a disorder. When faced with the uncertainty about whether the parent's or child's perspective is most accurate [32], several authors argue that the best practice is to combine information from both respondents in order to perform a better assessment [9,12,28].

Many children are aware of their stuttering shortly after onset [5, 32]. As they grow older, children may experience numerous situations and face many challenges that increase their awareness and concern about stuttering and may influence the way they see themselves. Many children who stutter perceive themselves as ineffective speakers; they may also have negative reactions and thoughts about themselves and their speech [21, 32, 33, 34] . Their reactions to stuttering are influenced by the ways in which stuttering affects them [9, 21, 32]; this may be observed as regards the performance, health, social life and occupation (e.g. [16,33,36,37]). Parents may perceive this impact in terms of struggle behaviours, as children attempt to convey messages, or in terms of emotional reactions, such as confusion, anxiety, despair, frustration, anger, and sadness [23]. Often, these emotional reactions can be associated with avoidance behaviours, such as withdrawal from play, reduced speaking, and asking others to speak instead [23]. Parents may, therefore, perceive the impact of stuttering on children's quality of life based on the difficulty their child experiences when communicating freely [23].

Parents often express concerns about their children who stutter. They report a lack of knowledge about what to do in order to help their children cope effectively with stuttering. In turn, parents also experience negative feelings and thoughts, such as guilt, anxiety, helplessness, distress, despair, sadness, emotional strain, fear, and lack of confidence in their skills [8, 38, 14, 21, 35, 36, 39].The parental concerns often relate to

fears about the consequences that stuttering may have on their children's future [9,23,40], such as difficulties regarding performance at school, struggles in peer relationships, and speech-motivated teasing or bullying [21, 35, 37].

Although the previously mentioned studies have explored the concerns of parents about their children who stutter [9, 21, 24,39], little is known about which factors are likely to influence the degree of concern. Attitudes, opinions, preconceptions and stereotypes about a concept result from an individual's own mental processes as well as from how new information is integrated as the person experiences different situations [42, 43]. Thus, earlier experiences with stuttering, such as having other family members who stutter or being people who stutter themselves, may influence parents' perceptions about stuttering and, consequently, affect how they interact with their children. Given the genetic aspects of stuttering [3], it is relatively common for parents of children who stutter to have personal experience with their own or a family members' stuttering. This could result in increased concern about daily situations [41, 43] such as that taking place at school, where negative experiences, due to speech, are pretty common [44].

Parental concerns can correspond to the actual experiences of the child and be consistent with children's responses or behaviours; however, some parents may not be fully aware of the difficulties that children face [23,45]. Cultural issues may also influence their perceptions due to culturally rooted stereotypes, beliefs, and attitudes toward stuttering [46, 48] . For example, research using the Public Opinion Survey of Human Attributes-Stuttering (POSHA-S) [48] has shown some differences among countries and cultures in how different people react to stuttering[49]. For example, it was found more negative attitudes in Italy and more positive in Norway and Sweden, compared to POSHA-S median values. Public stuttering attitudes in countries such as Germany, Poland, and Portugal were found to be closest to the median values [49].

Previous research studies showed that children who stutter perceive themselves as poorer speakers with a negative impact on their lives [9, 52, 56], in addition parents often succumb to negative emotions due to their children's speech [21, 37, 51, 52, 53]. Currently, little is known about whether or not parents and children are in agreement with one another about how they perceive the impact of stuttering. Therefore, the main goal of this study is to explore parents' perceptions about stuttering and to compare it with children's perceptions by using the same measure, to allow share the parameters analysis for a better knowledge of stuttering impact views.

2. Methods

2.1 Participants

Participants were 50 children aged 7 to 12 years old (M= 9.10 years old; SD=1.7 years) and 50 parents aged 31 to 55 (M= 42.26 years old; SD =4.823).

The parents provided information about their socio-demographic background and the child's stuttering via a checklist created for this study. The questionnaire gathered information about the participants' sex, history of prior therapy, education level, and family history of stuttering.

The majority of parent respondents were mothers (n=44); the respondents had a mean overall education level of bachelor's degree (n=32). Table 1 shows the demographic characteristics, such as age, sex, and education level, for both parents and their children.

The sex ratio of children who stutter was 2.6 boys to each girl (36 male and 14 female). This figure is generally consistent with previously reported values [6, 53] . The sample includes only children without any neurological impairment, psychiatric disturbance or history of head injury, learning disorder or seizure. Participants were recruited from speech-language therapist cases and through referral of teachers and other professionals throughout Portugal. Of the 50 child participants, 50% were in therapy at the time of the study, 22% had received therapy previously and 28% were waiting for therapy or just initiating therapy. The children who were in therapy at the time of data collection had been in treatment between 1 month and 96 months (M= 9.30 mos.; SD = 19.38 mos.). Children who had previous therapy had received treatment between 3 months and 48 months (M=13.28 mos.; SD = 12.99 mos.). Concerning previous family history of stuttering, 30 of the children had a family history of stuttering, whereas 20 did not.

Table 1: Demographic characteristics of the children (N=50) and parents (N=50)

Children						
Group	Family history of stuttering (n=30)		No family history of stuttering (n=20)		Total	
Age mean (SD)	9.20 (1.75)		8.95 (1.73)		9.10 (1.70)	
Sex (M/F)	21/9		15/5		36/14	
Education Level (n)						
1st grade	6		2		8	
2nd grade	5		6		11	
3rd grade	6		3		9	
4th grade	5		2		7	
5th to 7th grade	8		7		15	
Speech Therapy (n)						
Yes	15		10		25	
No	8		6		14	
Previous	7		4		11	
Parents						
Age mean (SD)	42.17 (3.95)		42.40 (6.01)		42.26 (4.82)	
Sex (M/F)	2/28		4/16		6/44	
Education Level (n)	Mother	Father	Mother	Father	Mother	Father
7-9 years	2	1	3	5	5	6
10-12 years	6	8	2	2	8	10
Bachelor's degree	19	18	13	12	32	30
Master	1	3	2	1	3	4
Phd	2	0	0	0	2	0

2.2 Materials

The SSI-4 [55] was used to confirm and diagnose stuttering, along with the Portuguese story, "*A história do rato Artur*" [56]. This story has been used in several Portuguese studies (e.g. [57, 58]) because it has a high test-retest consistency, enabling the comparison of two temporal moments for the same speaker, and happens to be a phonetically-balanced text, which has been interpreted as indicating that it is close to spontaneous discourse [59]. Some of the younger children found it hard to read the story (N = 8), which is why only the SSI4 plates were used for them.

The European Portuguese version of *Overall Assessment of the Speaker's Experience of Stuttering for School-Age Children, age 7-12* (Avaliação Global da Experiência Subjetiva da Gaguez: OASES-S-PT) [60] was used to explore children's perception of stuttering. The OASES-S is divided in 4 sections: a) Section I (General Information), contains 15 items pertaining to the speakers' perceived fluency and speech naturalness, knowledge about stuttering, and overall feelings about stuttering; b) Section II (Your Reactions to Stuttering), contains 20 items examining the speakers' affective, behavioural, and cognitive reactions to stuttering; c) Section III (Communication in Daily Situations), contains 15 items assessing speaker degree of difficulty when communicating in general situations, at school, in social contexts, and at home; d) Section IV (Quality of Life), contains 10 items assessing how much stuttering interferes with the speakers' satisfaction with their ability to communicate, their ability to participate actively in life, and their overall sense of well-being.

Parent perceptions of the impact of stuttering were gathered via a parents-adapted OASES-S version that was developed by the authors and a panel of experts. The authors made the first adaptation through OASES-S-PT and the panel of experts made a revision according to delphi methodology. The OASES-S-PT-P (*Avaliação Global da Experiência Subjetiva da Gaguez - idade escolar - Versão Portuguesa – Adaptação para pais*) [61] includes the same sections and items as the OASES-S, though it was modified so that parents are questioned about their children. For example, the question: "*How often can you speak fluently (without stuttering)*" was turned into: "*How often can your child speak fluently (without stuttering)?*"

2.3Procedures

Collecting children data took place at the same time as with parent data; the socio-demographic checklists and OASES-S-PT–P were completed by parents while, in a different quiet room, the researcher assessed the child with the OASES-S-PT and SSI-4. All testing was conducted between December 2017 and June 2018.

Data analysis was conducted with SPSS (Statistical Package for Social Sciences - Version 24 for Windows). The Kolmogorov-Smirnov test was performed in order to determine the normality of OASES scores for Portuguese parents and children. Results of the statistical test showed normal distributions for some OASES scores and non-normal distributions for other OASES scores.

To explore the relevance of having family history of stuttering, the participants were grouped taking into account this factor: group with family history of stuttering (n=30 children and n=30 parents) and group without family history of the disorder (n=20 children and n= 20 parents). When comparing mean scores of children and parents with and without family history of stuttering, paired-samples t-tests were used for normal distributions and Wilcoxon Signed-Ranks tests were used for non-normal distributions.

3. Results

Regarding both children and parents scores, Figure 1 show the impact ratings for OASES-S-PT sections and for the overall impact rating. Most of the children (52%) reported a moderate overall stuttering impact rating. Approximately half of the parents (44%) rated their children as experiencing a mild-to-moderate overall stuttering impact. None of the parents rated their children as experiencing a severely adverse impact as a result of their stuttering. The same is true for children perceptions. Scores were higher for section I (General Information), regarding both children and parents, and they were lower for Section IV (Quality of Life), regarding the children group, and for Section III, regarding the parents' group.

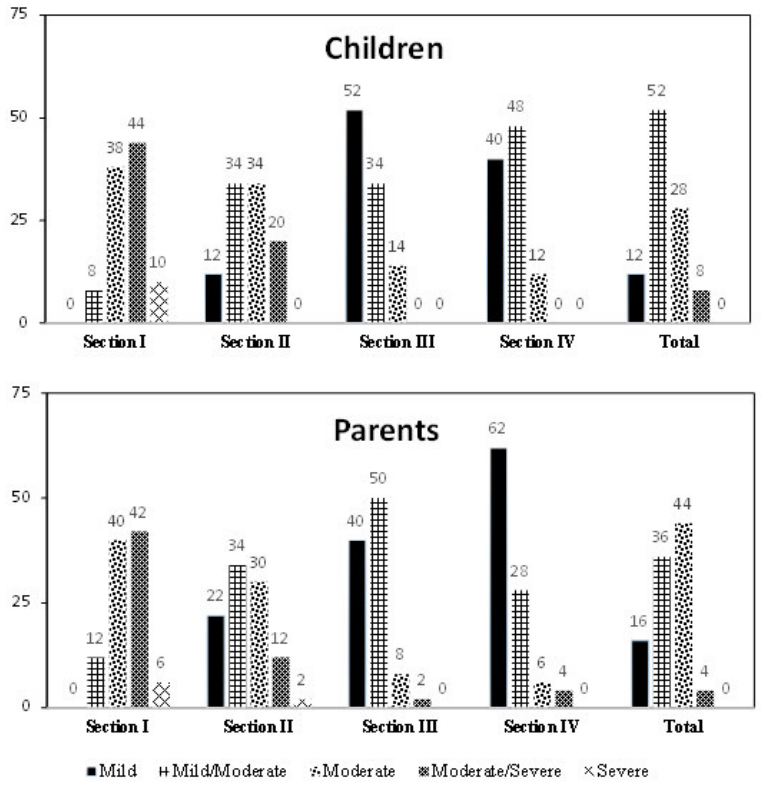

Fig 1. Impact ratings by children and parents

Table 2 shows the mean rating scores for both children and parents. For children, mean scores ranged between 1.50 and 2.89, with lower mean scores for Section IV in both groups: children with family history of stuttering (Quality of Life; M=1.50; SD=0.58), and children without family history of stuttering (Quality of Life; M=1.59; SD=0.71); and higher mean scores for Section I in children with family history (General Information; M=2.87; SD=0.64)., and in children with no family history of stuttering (General Information; M=2.89; SD=0.71). For parents, mean scores ranged between 1.44 and 3.01, with lower mean scores for Section III (Communication in Daily Situations; M= 1.44; SD=0.42) and higher mean scores for Section I (General Information; M=3.02; SD=.61), both in the group with family history of stuttering and in the group with no such family history.

When comparing the mean impact rating scores between children who stutter and their parents, statistically significant results were observed as regards the family history of stuttering in the group with higher mean scores for parents in the Section II (Reactions to Stuttering*:* $t_{(29)} = 2.90$, $p = .01$), Section IV (the Quality of Life: $z = 2.20$, $p = .03$) and overall score ($t_{(23)} = -2.25$, $p = .03$).

Table 2: *Mean (M), standard deviations (SD) and p-values for the impact scores of children who stutter (n=50) and their parents (n=50)*

	Family history of stuttering (n=60)				No family history of stuttering (n=40)			
Section Scores	Children M (SD)	Parents M (SD)	t/z	*p*	Children M (SD)	Parents M (SD)	t/z	*p*
I	2.87 (.64)	3.03 (.48)	t=-1.60	.12	2.89 (.71)	3.02 (.61)	t=1.09	.29
II	2.07 (.67)	2.51 (.72)	t=-2.90	**.01****	2.28 (.92)	2.05 (.63)	t=1.30	.21
III	1.56 (.40)	1.61 (.51)	t=.57	.57	1.64 (.65)	1.44 (.42)	t=1.88	.08
IV	1.50 (.58)	2.25 (.45)	z=-2.20	**.03***	1.59 (.71)	1.96 (.47)	z=-.24	.81
Overall	2.04 (.71)	2.25 (.61)	t=-2.25	**.03***	2.15 (.62)	1.96 (.47)	t=1.88	.08

*$p < .05$, **$p < .01$

Note: I -General Information; II - Reactions to Stuttering; III - Communication in Daily Situations; IV- Quality of Life

4. Discussion

This study compared children and parents' views on the impact of stuttering on children's lives using an instrument with the same domain structure. Analyses revealed that parents are generally aware of the impact of stuttering on their children's lives, however, they also showed interesting differences. These were particularly apparent in

the group of children with a family history of stuttering, especially reflecting both perceptions of the children's reaction to stuttering, and its impact on their quality of life.

Portuguese children who stutter end up suffering an overall moderately-adverse stutter-related impact. This was particularly evident in Section I (General Information), which revealed that children have a low overall knowledge or self-awareness of stuttering . In addition, parents' responses in Section I revealed that they think that their children have little knowledge about stuttering and may find it hard to accept the disorder. Considering that most of the children either had received or were receiving therapy, this was unexpected, for it was predictable that the children would have greater knowledge about the disorder. It is possible that parents' responses reflected their own lack of knowledge and this would be consistent with studies reporting that parents have a general lack of knowledge about stuttering, and about what they can do to help their children deal with stuttering effectively [9,15,23,40]. Such findings highlight the need for Portuguese therapy programs to provide more information and education about stuttering, not only for children, but also for parents [15,37,40,62]. Parents' feelings of guilt, helplessness, and worry might be mitigated if they had more knowledge about the disorder, and this might help them to develop more confidence and ability when it comes to supporting their children [14, 62].

In Section II (Reactions to Stuttering), parents' and children's responses are in agreement with previous literature which reports greater negative emotional reactions, including embarrassment, frustration, and anxiety in children who stutter [9 , 52, 63, 64]. Responses of both groups show that children were experiencing negative emotions associated with their stuttering.

In Section III (Communication in Daily Situations), children reported being able to communicate freely in several scenarios, such as at school, at home, and in social settings; however, it was clear that some situations were found harder than others, such as speaking in large groups. Parents also reported their perceptions of their children's communication difficulties and indicated that some situations were found harder than others, particularly, speaking in large groups, giving a presentation or speaking in front of the class, asking a question, and read aloud to the class. These results are in agreement with other reports regarding parent concerns about their children's performance at school [9,23,40].

Children's responses on Section IV (Quality of Life) revealed that Portuguese children experience a minimal impact associated to stuttering. At the same time,

parents' responses indicated a perception that stuttering may have a mild to moderate impact on children's quality of life. This was particularly evident in parents' ratings concerning the impact of stuttering on life at large, the reaction of other people to stuttering (e.g. teasing), and difficulties in attending social events (e.g. sports teams, sleeping at friends' houses, parties, among others). Again, these results are concordant with studies about how stuttering impacts quality of life and with studies about parental concerns [23,36–39, 40, 41, 44]. Children's results may be due to a less-than-perfect understanding of the issues in this section (as compared to other sections).

Results from this study highlight the need to increase children's and parents knowledge about stuttering. Giving knowledge is likely to increase their confidence in managing stuttering and minimize negative reactions about their communication abilities. Also, findings suggest the need to include in Portuguese programs strategies to communicate in specific scenarios or contexts, especially with peers and in large groups, as well as the need for a partnership between clinicians and teachers [45]. Finally, findings highlight the need for Portuguese therapy programs to increasingly aim at reducing negative reactions to stuttering, for both parents and children [15,37,40,62].

A comparison of parent impact scores for participants with a family history of stuttering versus participants with no family history of stuttering revealed significant differences: in those families with a history of stuttering, parents perceive a greater impact of stuttering in the items related to quality of life and children's reactions to stuttering. These results are in agreement with Vanryckeghem study (1995) who didn't find a significant parent-child agreement comparing scores from both froups in CAT-D [24] and suggest that having prior experience with stuttering through a family member could be a contributing factor that increases parental concerns. All these concerns, especially regarding a child's future [8, 36, 39] can be exacerbated when family members have had or still have limitations due to stuttering. In addition, parents in families with a history of stuttering may be more alert and more aware of stuttering because they have already faced with difficulties that stuttering brings. Results are in agreement with literature reporting the importance of previous experiences in generating attitudes, opinions, preconceptions, and stereotypes regarding a concept, such as a disorder [41, 43]. It may, therefore, be appropriate for clinicians to consider this factor to ensure that the parents' concerns about stuttering do not help increase the children's concerns.

The responses of both groups concerning items related to general information and how children communicate in daily situations were mostly shared. These results reinforce the idea that parent self-report can be a valid measure for analysing certain domains of stuttering disorder. These results are in agreement with the studies conveying the underlying veracity contained in the information collected from parents [26- 30].

Main results reinforce just how important it is to include parents in the assessment, since their feedback about the impact of stuttering in their children‘s lives adds more value to both perceptions. Information provided by parents also provides clues about how concerned they are about their children’s future and about how this concern may influence family dynamics.

The present study has some methodological limitations that could bias the results. For example, the validity of the modified OASES-S-PT used in this study has not been verified. This study’s findings motivate a further consideration of children’s and their parents’ reactions to stuttering. Future research should also consider potential differences between children who have received treatment and children who have not (yet) received treatment. Although treatment status was collected in this study, details about the nature of that treatment were not collected. Therefore, an appropriate comparison between these subgroups could not be completed. Other important paths to follow in future research include comparing mothers’ and fathers’ perceptions and with other caregivers as well as correlate results with other variables, such as participant’s history of therapy.

5. Conclusion

In summary, this study delves into how parents of school-age children perceive the impact of stuttering on their children's lives and whether their perspective was consistent with their children’s. Findings highlight ways in which parents’ and children’s perceptions are consistent (General Information and Communication in Daily Situations) and ways in which their perceptions differ (Reactions to stuttering and Quality of Life), depending on whether there is a family history of stuttering or not. Findings also highlight the fact that parents and their children could have their own separate needs, feelings, thoughts, and concerns about stuttering that should be taken

into account when developing therapeutic programs. Therefore, children and their families should have customized therapy tailored to their specific fluency needs.

Parents can provide useful information about their children and about themselves that can help clinicians better understand the impact of stuttering on children's lives. This study reinforces the need to provide parents with information about stuttering. By investing people with knowledge about the disorder (in different contexts of children's life), they may end up feeling more confident managing stuttering, and they can learn how to adequately react to the child's disfluencies.

Understanding the differences between parents' and children's perspectives about stuttering can also help clinicians develop support programs for parents, as parents' worries and fears about children's future may influence how the disorder develops. Thus, successful approaches would be those that take into account, and support both children who stutter and their parents.

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CHAPTER 5: How do teachers perceive the impact of stuttering on school-age children?[5]

[5] The following study is under review and awaiting for acceptance: Rocha, M.S., Yaruss J.S. & Rato, J.R. How do teachers perceive the impact of stuttering on school-age children? *Revista Portuguesa de Educação.*

Abstract

Stuttering is a neurodevelopmental disorder involving interruptions in the natural flow of speech. The reactions of listeners and others in a child's environment could affect how children perceive their own stuttering. Because children who stutter experience many of their everyday social situations in the school context, it is essential to know how teachers perceive the impact of stuttering on their students. In this study, we collected data about teachers' perceptions of the impact of stuttering on Portuguese children who stutter using an adaptation of European Portuguese translation of Overall Assessment of the Speaker's Experience of Stuttering (OASES-S-PT). Participants were 27 teachers and their students who stutter (mean age=9.0 mos., SD=1.8 mos.), who were recruited from different cities in Portugal. Overall, teachers perceived the overall impact of stuttering in theirs students' lives as mild-to-moderate. Our results did not reveal any statistically significant differences between the teachers' perceptions about the impact of stuttering and the students' own self-reports. Still, there was no correlation between the students' impact scores and the teachers' impact scores, and teachers were unable to rate several items on the instrument. This indicates that teachers were unfamiliar with some aspects of the children's experiences with stuttering, which highlights the need to better integrate teachers into therapy programs to increase support within the environment of children who stutter.

1. Introduction

Stuttering involves more than just observable behaviors, such as repetitions, prolongations, and blocks (Bloodstein & Ratner, 2008). Instead, it should be viewed a multifactorial issue involving environmental, genetic and constitutional factors (Ambrose & Yairi, 1999; Conture, 2001; Smith & Weber, 2017) that can lead to activity and participation limitations in a person's life (Yaruss & Quesal, 2004b, 2006). Stuttering is likely to have an adverse impact on education, health, personal relationships, social life, and occupation (e.g. Beilby, Byrnes, & Yaruss, 2012; Boey, 2012; Craig, Blumgart, & Tran, 2009; Author 1, Author 2, & Author 3, 2019a; Yaruss & Quesal, 2016). Because of the broad-based nature of stuttering, it is important to take into account the perceptions and experiences of those who stutter when planning evaluations and treatment (Yaruss & Quesal, 2004a).

A recent study used the European Portuguese translation of the Overall Assessment of the Experience of Stuttering (OASES-S-PT, Author 1 et al., 2019b) to examine the experiences of Portuguese school-age children who stutter. Analyses revealed a mild-to-moderate overall impact, including low overall self-awareness of stuttering and negative emotional reactions associated with stuttering. Findings also revealed difficult in communicating in specific situations, such as speaking in large groups (Author 1 et al., 2019b).

For many children who stutter, the school-age years can be difficult due the need to engage in speaking activities associated with their education. These activities might include raising their hands to speak in class, asking and answering questions, and talking with teachers (Cooke & Millard, 2018; Daniels, Gabel, & Hughes, 2012). Adults who stutter have reflected on the difficulties they have experienced at school, especially when participating in speech-related activities (Klompas & Ross, 2004). Considering that children spend a lot of time in schools and that teachers are the primary adults with whom children interact in that setting, it is sensible to explore whether teachers can provide clinicians with relevant information about children's behaviors and affective reactions to stuttering (Boey et al., 2009; Bothe & Richardson, 2011; Langevin, Packman, & Onslow, 2010; Pros, Tarrida, Muntada, & Martin, 2017; Seixas, Matos, Festas, & Fernandes, 2014).

Several studies have shown that it is important for teachers to cooperate in the assessment and treatment of children with various conditions (Al-Awad & Sonuga-Barke, 2002; Firmin, Proemmel, & Hwang, 2005; Harlen, 2005; Schatz, Ballantyne, & Trauner, 2001; Ulloa, Narváez, Arroyo, del Bosque, & de la Peña, 2009). Analyses of the teachers' responses in these studies generally show that the information provided by teachers is valid; however, further information is needed regarding teachers' perceptions about stuttering to determine whether they are able to provide accurate information. If teachers are to serve as allies in the therapy process, it is important that they have appropriate knowledge about stuttering, so that the information they share can be useful for parents and clinicians (Blood, Boyle, Blood & Nalesnik, 2010; Cooke & Millard, 2018; Hayhow, Cray, & Enderby, 2002). Therefore, the purpose of this study was to explore teachers' perceptions about the impact of stuttering on school-age children who stutter.

2. Methods

2.1 Participants

Participants were 27 teachers and their students who stutter (mean age=9.0 mos., SD=1.8 mos.), who were recruited from different cities in Portugal. The majority of the teacher participants were women (2 male and 25 female).

Table 1 shows: demographic characteristics of teachers, general information about the teachers' level of experience, and information about the teachers' experiences with stuttering and with students who stutter. On average, teachers reported knowing their students who stutter for more than one year (M= 13.6 mos.; SD =13.3 mos.), with a minimum of 2 months and a maximum of 48 months.

Of the teacher participants, 55.6% reported that they had not previously had a student who stuttered in their class. Interestingly, 40.7% reported that they had never talked about stuttering with their students, though 48.1% indicated that they had discussed stuttering with their students' parents. The majority of participants reported lacking knowledge about stuttering (63.0%). Of the teachers who claimed to have some knowledge about stuttering, 18.5% reported that they acquired this knowledge through an online search.

Table 1: Demographic characteristics of the teachers (n=27)

Teachers variables	M (SD)	
Teaching in schools (years)	20.63 (9.50)	
Teaching children who stutter (months)	13.56 (13.28)	
Number of students who stutter	1.87 (0.83)	
	n (%)	
Sex	Male	Female
	2	25
Previous experience teaching students who stutter	Yes	No
	12 (44.44%)	15 (55.56%)
Stuttering conversation	Parents	Students
Just once	2 (7.41%)	1 (3.70%)
Frequently	6 (22.23%)	2 (7.41%)
Never	5 (18.52%)	11 (40.74%)
Sometimes	13 (48.14%)	8 (29.64%)
Rarely	1 (3.70 %)	4 (14.81%)
Non answered	-	1 (3.70%)
Classroom interference	Yes	No
	2 (7.41%)	25 (92.59%)
Stuttering knowledge		
No	17 (62.96%)	
Academic education	2 (7.41%)	
Online search	5 (18.52%)	
Workshop and training	2 (7.41%)	
Other	1 (3.70%)	

Table 2 shows demographic characteristics about the children's age, sex, education level, and prior history therapy. The sex ratio of children who stutter was 3.5 boys to each girl (21 male and 6 female). This figure is generally consistent with previously reported values (Craig, Hancock, Tran, Craig, & Peters, 2006; Yairi & Ambrose, 2005).

The sample includes only children without any neurological impairment (other than stuttering), psychiatric disturbance, or history of head injury, learning disorder, or seizure disorder. Participants were recruited from speech-language therapist caseloads and through referral of teachers and other professionals throughout Portugal. The sample was taken from a broader study (n=50) about the impact of stuttering on

children's lives (Author 1 et al., 2019b). Of these, 27 children were selected for having teacher data, and, in which, 44.4% were in therapy at the time of the study, 29.6% had received therapy previously, and 25.9% were waiting for therapy or initiating therapy. The children undergoing therapy at the time of data collection had been in treatment between 3 month to 16 months (M= 14.8 mos.; SD = 28.8 mos.). Children who had undergone previous therapy had received 3 to 48 months of treatment (M=13.3 mos.; SD = 14.6 mos.).

Table 2: Demographic characteristics of the children (n=27)

Children variables	n (%)	
Sex	Boys	Girls
	21(77.8%)	6 (22.2%)
Education level		
1st grade	6 (22.2 %)	
2nd grade	6 (22.2 %)	
3rd grade	4 (14.8%)	
4th grade	4 (14.8%)	
5th to 7th grade	7 (26%)	
Speech Therapy		
Yes	12 (44%)	
No	8 (25.9%)	
Previous	7 (29.6%)	

2.2 Materials

The SSI-4 (Riley, 2009) was used to confirm and diagnose stuttering, along with the Portuguese story, "*A história do rato Artur*" (Guimarães, 2007). This story has been used in several Portuguese studies (Guimarães & Abberton, 2005; Silvestre, 2009) because it has a high test-retest consistency and is phonetically balanced. This has been interpreted as indicating that it is close to spontaneous discourse (Moon, Chung, Park, & Kim, 2012). Some of the younger children found it hard to read the story (n = 8), so only the SSI4 plates were used for them.

Tables 1 and 2 provide information about teachers and their students collected through a checklist created for this study. Parents provided information about their children's stuttering and socio-demographic background, including sex, therapy history, education level, and family history of stuttering, via another checklist that was also created for the purpose of this study.

The European Portuguese version (Author 1 et al., 2019a) of Overall Assessment of the Speaker's Experience of Stuttering (OASES-S; Yaruss & Quesal, 2016) was used to explore how the child participants in this study were impacted by their stuttering. An adaptation of the OASES specifically for teachers (OASES-S-PT-T) was created to explore teachers' perception of how stuttering impacts their students' lives.

The OASES-S is divided into 4 sections: Section I (General Information) contains 15 items pertaining to the speakers' perceived fluency, speech naturalness, knowledge about stuttering, and overall feelings about stuttering; Section II (Your Reactions to Stuttering) contains 20 items aimed at examining speakers' affective, behavioral, and cognitive reactions to stuttering; Section III (Communication in Daily Situations) contains 15 items aimed at assessing how hard speakers find it to communicate in generic scenarios, at school, socially, and at home; and Section IV (Quality of Life) contains 10 items aimed at assessing how much stuttering interferes with speakers' satisfaction with their ability to communicate, their ability to actively participate in life, and their overall sense of well-being. The OASES-S-PT-T also includes 4 sections, items similar to the ones found in OASES-S. Slight changes were made to verb conjugations and pronouns so the questions were directed to teachers. For example, the first question: "How often can you speak fluently?" was modified into: "how often is your student able to speak fluently (without stuttering)?"

2.3 Procedures

This study received full approval by the Ethics Committee of the Institute of Health Sciences of Universidade Católica Portuguesa (register number 34/2017). Prior to their participation in this study, parents provided informed consent for themselves and their children. Consent also included permission for the researcher to record the child and participants' right to withdraw from the study at any time was clarified. The teachers agreed to participate in study with the permission of the school board.

Assessment of the children and collection of parent-provided information were carried out simultaneously in different rooms. Sociodemographic checklists were filled out by parents (about 5 minutes) while, in a different quiet room, the researcher performed the SSI-4 with the child (about 15 minutes).

The OASES-S-PT-T and the teachers' checklist were provided to the teachers via the parents. The forms were placed in a sealed envelope, together with a cover letter explaining the aims of the study. Once completed by the teachers, the documents were returned to parents who then forwarded them back to the researchers.

All testing was conducted between December 2017 and June 2018.

2.4 Data analysis

Data analysis was conducted with SPSS (Statistical Package for Social Sciences - Version 24 for Windows). The Kolmogorov-Smirnov test was performed in order to determine the normality of impact scores for Portuguese teachers and children. In the comparison of scores between children and teachers, paired-sample t-tests were used for variables with normal distributions, and Wilcoxon Signed Ranks tests were used for variables with non-normal distributions. Correlations between the children's impact scores and the teachers' impact scores were evaluated using Spearman's rho.

3. Results

3.1 OASES-S-PT-T scores for teachers of European Portuguese children who stutter

Figure 1 shows the distribution of scores for all four sections and overall impact ratings for both children and teachers.

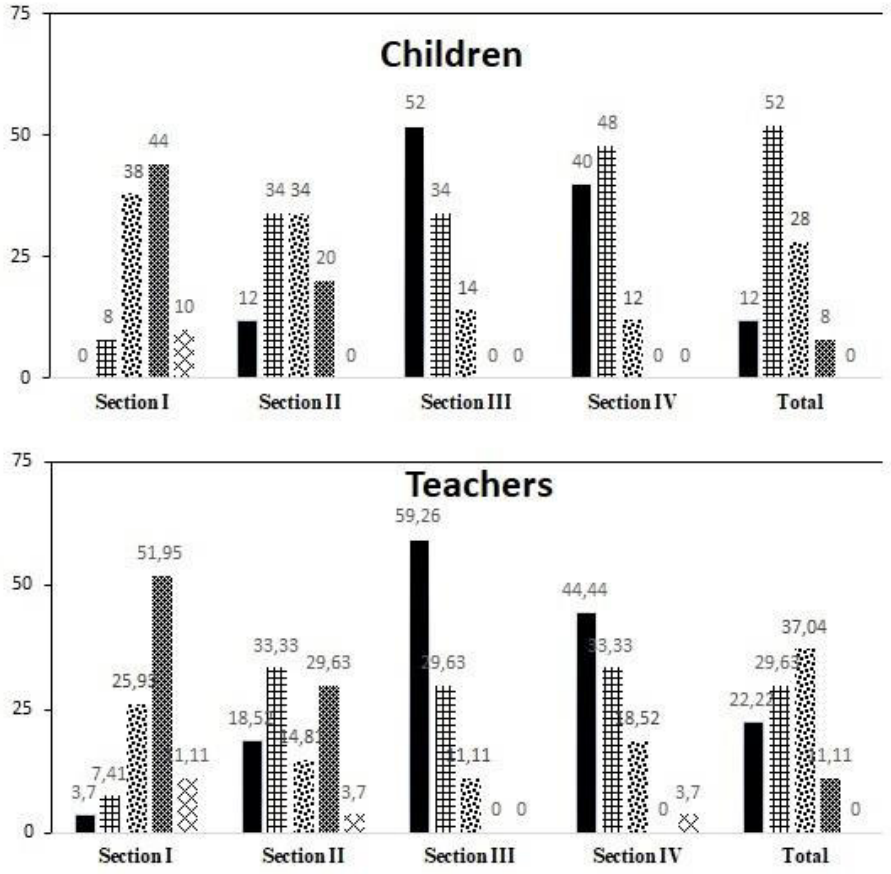

Figure 1: Impact ratings by children and teachers

In the group of Portuguese teachers, 37.04% indicated their belief that their students experienced a moderate overall impact due to their stuttering; 29.6% reported a mild-to-moderate impact, 22% reported a mild impact; and 11.1% reported a moderate-to-severe impact. None of the teachers in this sample rated their students as experiencing severe impact of stuttering in their life.

Examining the individual sections of the OASES revealed higher negative impact in Section I (General Information), with about half (51.9%) of the teachers rating the impact of stuttering in their student's lives as moderate-to-severe. Less impact was revealed in Section III (Communication in Daily Situations) and Section IV (Quality of Life), with approximately half of Portuguese teachers rating their students with a mild impact (50.3% and 44%, respectively). In Section II (Their Reactions to Stuttering), 33.3% of teachers rated their students as experiencing a mild-to-moderate impact; and 29.6% reported a moderate-to-severe impact.

The non-response rate for certain items was high, suggesting that teachers did not have insights into their students' experiences. Section I (General Information) had the highest rate of non-answers. Almost half of the participants (44.4%) failed to respond to questions about students' use of techniques or strategies learned in speech therapy. In Section II (Their Reactions to Stuttering), the least-answered questions (18.8%) addressed physical tension that the students might experience in their speech. In Section III (Communication in Daily Situations), teachers did not answer questions about the difficulties children experienced when communicating at home (18.5% to 29.6%). In Section IV (Quality of Life), 33.3% of teachers did not respond to the question about how much their students' lives were affected by needing to go to speech therapy.

3.2 Comparison and correlation of OASES-S scores of teachers and children

Table 3 shows the results of mean impact rating scores comparison between children who stutter and their teachers. Student's scores were obtained through another study (Author 1 et al., 2019b) and used for comparison purposes. There were no statistically significant between-group differences ($p > .05$) in the impact score means.

Table 3: Mean (M), standard deviations (SD) and p-values for the impact scores of children who stutter (n=27) and their teachers (n=27)

Impact Scores	Correlation coefficient (*p* value)
I	.200 (.317)
II	.215 (.281)
III	.319 (.105)
IV	.214 (.283)
Total score	.265 (.182)

As shown in Table 4, there was no significant correlation between students' impact scores teachers' impact scores ($p > .05$).

Table 4: Correlations (Spearman's rho) among student's impact scores and teacher's impact scores

Impact Scores	Children who stutter M (SD)	Teachers M (SD)	*t/z*	*p*
I	2.789 (.627)	3.033 (.688)	t= -1.531	.138
II	2.106 (.835)	2.318 (.924)	t= -.972	.340
III	1.495 (.430)	1.428 (.560)	z= -.745	.456
IV	1.540 (.604)	1.704 (.714)	z= -1.090	.276
Total score	2.022 (.542)	2.116 (.635)	t= -.664	.513

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4. Discussion

This study asked teachers about how stuttering impacts their students' lives and compared their responses to responses previously collected from the children. On average, teachers perceived a moderate overall adverse impact of stuttering, sharing the same rate with children.

According to the teachers, their students have little knowledge about stuttering, and these results are in agreement with previous studies (Author 1 et al, 2019b). Notwithstanding, teachers' also may be influenced by they own lack of knowledge about stuttering (Carroll, 2010; Li & Arnold, 2015; Silva et al., 2016). The large number of skipped answers to certain issues reinforces this notion.

Results suggests that teachers perceive stuttering children's reactions to stuttering to be negative. These reactions include emotions and attitudes toward speaking and stuttering such as embarrassment, frustration, and anxiety. Such findings support the idea that treatment for school-age children should include multiple goals based on individual needs, focused not only on enhancing fluency, but also on other goals, such as the acceptance of stuttering, minimizing avoidance, and reducing negative emotions associated with stuttering (Cooke & Millard, 2018; Murphy, Yaruss & Quesal, 2007; Reardon-Reeves & Yaruss, 2013; Yaruss, Coleman, & Quesal, 2012). Reducing children's negative emotions lead to the development of better communicative tools to use daily, softening stuttering impact (Howard, 2013; Murphy et al., 2007; Reardon-Reeves & Yaruss, 2013; Yaruss et al., 2012).

Teachers were able to identify adverse impact from stuttering in the school setting. These results are in agreement with previous literature about the stuttering impact on children, namely in academic performance (Boey, 2012; Beilby et al., 2012; Cooke & Millard, 2018; Craig et al., 2009; Guttormsen, Kefalianos, & Næss, 2015; Yaruss & Quesal, 2016). These results confirm the importance of gathering information from multiple actors in the child's life during the assessment process. In particular, teachers can provide unique insights into children's experiences in the classroom context, and this can lead to better cooperation between clinicians and teachers in the treatment of children who stutter (Blood et al., 2010; Cooke & Millard, 2018; Daniels et al., 2012; Miller, 1999; Murphy et al., 2007; Reardon-Reeves & Yaruss, 2013). Findings also highlight the need for treatment to address children's communication difficulties in specific scenarios, such as when speaking with peers and in large groups.

Teacher's perceptions of the impact of stuttering on children's lives may have been influenced by their own perceptions of stuttering, as well as by their views about the future for their students who stutter (Dorsey & Guenther, 2000; Turnbull, 2006). Teachers' negative stereotypes and perceptions regarding people who stutter have previously been identified (Dorsey & Guenther, 2000; Turnbull, 2006), and this might have contributed to teacher's perceptions of negative impact on their students' lives.

Although teachers were able to provide insights about children's experiences in the school setting, they – understandably – were unable to provide information about children's communication difficulties at home or in other settings outside of the school. This is made clear by the number of skipped items in the teachers' responses on the OASES. It may also reflect the teachers' lack of understanding about stuttering and a

lack of information sharing between speech therapists and teachers, since the majority of teachers did not know how to respond to treatment-related issues, stated themselves that they had no knowledge of stuttering and never talked about stuttering with their students. This is in agreement with studies that report a lack of teachers' knowledge about what they can do to help their students deal with stuttering (Li & Arnold, 2015; Silva et al., 2016). Moreover, the work of teachers with their students, regarding the demystification of the problem in the classroom, can also be fruitful.

Together, these findings emphasize the importance of incorporating teachers into the therapy process. The results highlight how fundamental it is to implement awareness-raising, teacher-targeted efforts. Brochures and videos may be used to provide general information, as well as holding regular meetings in which parents, teachers, and therapists would participate (Carroll, 2010).

In order to explore the differences between teachers' and their students' answers, a score mean comparison was carried out, which revealed no statistically significant results. This could mean that the responses of both groups were, in general, very similar. However, there was no significant correlation between the impact scores of students and teachers, suggesting that the teachers were not entirely in tune with their students' experiences with stuttering. The low correlation may be related to the number of missing items in the teachers' responses. Regardless, findings highlight the need to improve partnership among teachers and speech therapists, and increase teachers' knowledge about stuttering (Carroll, 2010; Li & Arnold, 2015).

4.1 Limitations and Future Directions

Findings should be interpreted with caution due to the relatively small sample. This study focused specifically in the perceptions of teachers; additional research will be needed to better understand whether there are differences in the perceptions of other professionals (such as speech-language pathologists), as well as siblings and other family members. In addition, cultural factors may influence teachers' perceptions, so the replication of this study in other countries will be important in order to customize therapy programs according to each country's cultural identity.

4.2 Clinical Implications

The results suggest the need to develop a closer and more effective relationship between therapists and teachers, especially because the perceptions about stuttering may have an impact on the way in which teachers cope with students who stutter.

Present findings provide a starting point for developing outreach campaigns that can help to demystify stuttering for teachers. Although teachers in this study did perceive that their students were experiencing negative impact as a result of stuttering, there were several areas in which the teachers did not have clear insights. This suggests that speech-language pathologists can do more related to stuttering education, about the experiences that are common for children who stutter, and, in particular, the nature of speech therapy for children who stutter (e.g., Reardon-Reeves & Yaruss, 2015).

5. Conclusion

This study highlights the importance of ensuring that teachers have an adequate understanding of the experiences of children who stutter. Although they may perceive the impact of stuttering in general, there are some areas where they are lacking insight. Improving teachers' understand of stuttering will help to create a supportive school atmosphere for children who stutter.

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Author 1, Author 2 &, Author 3 (2019b).

DISCUSSION

Research Aims and methodological aspects

The four studies detailed in this dissertation used the same sample of participants. The participants came from different parts of the country; therefore, the instruments were administered by the principal author of this work as well as by different Speech Language Therapists who were instructed to carefully follow the specified protocol. This protocol consisted of a document with an explanation of the study and guidance about how to administer each instrument used in the study (Appendix 1 and Appendix 2). Two versions of the protocol were prepared, each with the instruments in a different order of administration in order to reduce bias in the results related to order effect.

Although the participants in the study had previously been diagnosed as stuttering, the Stuttering Severity Instrument (SSI4; Riley, 2009) (Annex 3), with the Portuguese story, "*A história do rato Artur*" (Guimarães, 2002) (Annex 4) was used to confirm this diagnosis. In addition, in order to achieve the aims for the studies detailed above, all children were also assessed with the Portuguese versions of Multidimensional Anxiety Scale for Children (MASC) (Annex 5) and Children's Color Trails Test (CCTT (Annex 6). Children who stutter were also assessed with OASES-S-PT (Appendix 3). In order to protect copyright all Speech Language Therapists, which collaborated in the study signed a statement agreeing to the exclusive use of the instrument within the scope of the research work (Appendix 4).

The Sociodemographic checklists (Appendix 5-7), and the other questionnaires directed to parents and teachers were delivered in person or sent in a sealed envelope to the research collaborators to maintain confidentiality of responses. Beyond the checklist, questionnaires delivered to parents encompassed the Portuguese version (Appendix 8) of TMCQ (Annex 7) and the OASES-S-P (Appendix 9). A version of this questionnaire (OASES-S-T; Appendix 10) was also given to the teachers, along with the sociodemographic checklist and a brief letter presenting the research.

Prior to their participation in this study, all parents provided informed consent for themselves and their children. Consent also included permission to audio/video record the child (Appendix 11 and Appendix 12). The right to withdraw from the study

at any time was clarified. A cover letter was also prepared and delivered to the schools where sample collection was performed (Appendix 13).

This study received full approval by the Ethics Committee of the Institute of Health Sciences of Universidade Católica Portuguesa (register number 34/2017) (Appendix 14 and Appendix 15).

Achievement of Research Aims

This study primarily aimed to analyze the temperament, Executive Functioning (EF), and anxiety in children who stutter and to compare their performance with children who do not stutter. It also analyzed the impact of stuttering in the lives of Portuguese children who stutter, according to children's, parents', and teachers' views.

By studying temperament, EF, and anxiety, jointly, we added further data to multidimensional models of stuttering, such as Dual Diathesis – Stressor model (Choi et al., 2016; Walden et al., 2012). Our findings suggest that endogenous abilities, such as temperament and executive functioning, can play an important role in the development of stuttering. Children who stutter tended to be more reactive and sensitive compared to their nonstuttering peers. However, these results were not consistent across the age groups. Younger children in the group who stutter exhibited, in the executive functioning task, longer execution times, a higher number of warnings and failures, and a higher number of color sequencing errors compared to their nonstuttering peers. This suggests that children who stutter, in the first years of schooling, may have a tendency for attention weaknesses and impulsivity compared to their nonstuttering peers. Older children who stutter revealed on the temperament measure, comparisons between children who stutter and their nonstuttering peers revealed that older children who stutter exhibited significantly higher scores on the Anger/Frustration, Impulsivity, and Sadness subscales, and lower averages on the Attention/Focusing, Perceptual sensitivity, and Soothability/Falling reactivity subscales. Our results can be linked to emotional regulation, suggesting that children who stutter are more likely to be sensitive. This finding corroborates other studies that have explored temperament factors (Ambrose et al., 2015; Costelloe, Cavenagh, & Davis, 2015), and EF (e.g. Wolfe & Bell, 2004) in isolation. It also lends support to the DD-S model (e.g. Choi et al., 2016), which analyzes stuttering in a multidimensional view.

An important finding was the difference between temperament factors and EF abilities in the two age groups. This indicates that these cognitive and emotional

abilities are mediated by age and development. Again, this is consistent with a dynamic perspective on stuttering (e.g., Choi et al., 2016).

Endogenous abilities and exogenous contexts can play an important role as stuttering affects children's lives; this leads us to the last goal of this dissertation, related to the impact of stuttering on the lives of Portuguese children who stutter. In these studies, we examined impact as perceived by children, parents, and teachers' views.

To date, there was no published standardized measure for the assessment of the impact of stuttering in Portuguese children. Prior instruments have provided important guide about an anamnesis script for stuttering, as the *Protocolo de Anamnese de Gaguez*; *Perguntas específicas para a criança com gaguez* and *Protocolo de Avaliação da Severidade da Gaguez* (Valente, 2009)*;* however, the OASES-S-PT is the first validated measure that can be used for Portuguese children.

Using the OASES-S-PT, we found that Portuguese children who stutter experience a moderate overall impact of stuttering in their lives. More specifically, they experience a significant amount of negative impact on their communication abilities, as well as a low overall knowledge and self-awareness of stuttering. Compared to children in other countries, Portuguese children reported experiencing similar adverse impact to children in the Netherlands; however, Portuguese children who stutter reported less adverse impact than children in Western Australian, and less impact on daily communication than children in the USA, the Netherlands, and Western Australia. Even if it is not possible to make a direct and valid comparison between countries, due to some differences in sampling methods, the differences found reinforce the value in considering different cultural perceptions.

An important finding from this dissertation is related to the impact perceived by the people in a child's environment, in particular, the parents and teachers; results reinforce the need for these individuals to work in partnership to help children who stutter. Although parents are certainly important informants in the evaluation of the child who stutter, it is crucial to understand that their previous experiences, such as the presence of a family history of stuttering, can influence the ways they see the disorder. This in turn can influence their responses in self-reports. Moreover, our research makes it clear that teachers perceive stuttering children's reactions to stuttering to be negative; however, the relatively high number of skipped answers on the OASES-S-PT-T highlights the lack of insight that teachers have about the ways that stuttering affect

children's lives outside of the school setting. The results suggest the need to provide additional knowledge and education about the stuttering disorder in order to help teachers to better support their students who stutter.

Limitations

Our results should be interpreted in light of limitations that may have influenced the results. For example, participants who stutter were all recruited from the caseloads of cooperating speech-language pathologists. Some of the children were only at the evaluation stage, while others had almost completed therapy. The distribution between children who had already received treatment and children who were at the beginning stages of treatment was unbalanced. This may have contributed to differences between children in factors such as anxiety or awareness of stuttering, which may have been addressed in treatment (Davis, Bruce, & Gunnar, 2002). Moreover, we do not have a full understanding of the nature of treatment provided to children. Another example is seen in the ratio between boys and girls within the samples. For the group of children who stutter, the sex ratio was consistent with that found in the literature for other populations of people who stutter; the sex ratio for the nonstuttering children matched that of the general population. Because children who stutter and children who do not stutter were not matched by sex in this study, this contributed to an unbalanced sample that may have affected some of the between-group comparisons.

We also recognize limitations in this work in the use of self-report measures, in order to measure temperament, anxiety, and impact of stuttering. For example, in future studies, for anxiety measures it could be interesting to use speech tasks, rather than questionnaires, to trigger anxiety. Nevertheless, for the research on the impact of stuttering, despite using self-report measures we also used multiple informants, which increases the probability of collecting reliable information. It may be useful for the future studies to include observational measures in the assessment of anxiety and temperament features, as well as self-reports with multiple informants (e.g. parents and teachers). Furthermore, assuming that anxiety can arise as a secondary effect of living with stuttering, it could be interesting to use specific measures for different types of anxiety (e.g. state anxiety).

Clinical Implications

Our main findings suggest that stuttering treatment should be individualized and based on an assessment that includes emotional components, attitudes, and impact on life. The results from this research validate the OASES-S-PT and show the ways that this tool can contribute to a more effective assessment of the impact of stuttering on the lives of Portuguese children. Our results also emphasize the importance of combined direct treatment with children and approaches for working with parents, teachers, and other relevant people for the child. Findings also confirm that Portuguese school-age children who stutter can experience a moderate negative adverse because of stuttering. In order to refine Portuguese therapeutic programs, it will be important to include more components of desensitization and acceptance work in treatment.

Indirect treatment should be focused on environment monitoring through strategies that can be worked with parents. Because endogenous capabilities can be activated by contexts, it is critical for clinicians to have a detailed knowledge of internal factors and environmental factors for each child. Examples of environmental factors include daily routines, people's reactions to stuttering, and the demands to which the child is exposed. It is essential to work in partnership with parents and teachers, because they are the people who spend the most time with the children in this age group. Based on this dissertation finding, parents and teachers are generally aware of the impact of stuttering on the lives of children who stutter, so they can be reliable informants in the therapy process. Still, our results also highlighted the relevance of previous experiences as well as the lack of insight that teachers might have about some important issues related to stuttering, which reinforces reinforces the need to provide appropriate education about the disorder.

Future directions

Further studies along these lines, following a multidimensional view of stuttering, will contribute to a better understanding of the complexity of stuttering and support the development of individualized intervention programs. For example, studying the OASES in several countries may allow the creation of comprehensive, biopsychosocial, and personalized treatment program approach taking into account each person, and culture.

To our knowledge, this is the first work including normative data for Portuguese assessments in children who stutter. There is a need to develop additional studies

evaluating Portuguese children who stutter and Portuguese people who stutter in other age groups. Furthermore, because endogenous capacities, such as temperament and EF, can change over time, future research in this area should examine the interactions between temperament and EF over time. Additional research on the factors that contribute to the onset of anxiety should be carefully analyzed, at various age groups and related to endogenous abilities.

CONCLUSION

This dissertation reinforced the fact that stuttering is a multidimensional disorder and how stuttering can affect children's lives. The main conclusions confirm that stuttering should not be simply described as a speech disorder, for it clearly includes emotional and cognitive components that may have an impact on quality of life. Certainly, stuttering has genetic components, but the environment also plays an important role in the expression and experience of that underlying genetic predisposition.

Therefore, assessment and treatment should focus on multiple factors, rather than solely on speech mechanics. Clinicians should take into account the perspective from parents and teachers, because they are the ones who spend more time with children and can add useful information to the evaluation and therapeutic process. Inclusion of these individuals will also significantly contribute towards the entire treatment process.

Stuttering therapy must be able to reach beyond the therapy room. It should prepare children for different situations in their lives and guide parents and teachers to deal in a balanced and positive way with stuttering. The human mind is constantly reading and interpreting the environment around us. Thus, if we shift our perceptions, our beliefs, and the way we look at the disorder toward a more positive view, then we can influence the way one deals with stuttering. This will lead to positive repercussions on quality of life for children who stutter. This is particularly important for children, who are focused on absorbing messages from their environment and integrating the models of the people around them as they form their own beliefs and perceptions. Helping children process their emotions surrounding experiences like stuttering can reduce the likelihood that they will develop negative reactions. In particular, it will help parents and teachers to provide children with non-reactive, non-judgmental responses to stuttering that can help to mitigate the impact of stuttering on their lives. This mission cannot be achieved without an integrated approach that considers all contexts of a children's daily life.

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Annexes

Annexe 1

OASES™

Overall Assessment of the Speaker's Experience of Stuttering

OASES–S
Response Form
Ages 7–12

J. Scott Yaruss, PhD, CCC-SLP, BRS-FD
Craig E. Coleman, MA, CCC-SLP, BRS-FD
Robert W. Quesal, PhD, CCC-SLP, BRS-FD

Name: _____

Birth Date: _____/_____/_____ Test Date: _____/_____/_____

ID Number: _____ Age: _____ Grade: _____ Sex: ❑ M ❑ F

General Instructions:

This form includes four sections of questions that ask about your *current* experiences with your speech and stuttering. For each question, please circle the number that applies to you. Please answer every question. If a question does not apply to you, please check the box and move on to the next question. If you are not sure how to answer any of the questions, you may ask for help. An adult can read the test to you if you would like.

For Office Use Only

Instructions for Clinicians: Calculate Impact Scores for each of the four sections on the OASES–S by first summing the number of points in each section (A) and then counting the number of items completed in each section (B). Divide the total number of points (A) by the number of items completed (B) to obtain the Impact Score. Impact Scores range between 1.0 and 5.0. Using the Impact Scores on the left-hand side of the table, determine the Impact Rating for each section.

				Impact Rating				
Section	A Points	B Items Completed	A ÷ B = Impact Score	Mild 1.00–1.49	Mild/Moderate 1.50–2.24	Moderate 2.25–2.99	Moderate/Severe 3.00–3.74	Severe 3.75–5.00
I	_____	_____ (min = 13)*	_____	❑	❑	❑	❑	❑
II	_____	_____ (min = 18)	_____	❑	❑	❑	❑	❑
III	_____	_____ (min = 13)*	_____	❑	❑	❑	❑	❑
IV	_____	_____ (min = 8)*	_____	❑	❑	❑	❑	❑
Overall (Total)	_____	_____ (min = 52)*	_____	❑	❑	❑	❑	❑

*Note: Reduce this number by the number of items marked "Not Applicable," if any.

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8 9 10 11 12 A B C D E

Product Number 30416

Annexe 2

frontiers in Psychology

ORIGINAL RESEARCH
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Temperament, Executive Functioning, and Anxiety in School-Age Children Who Stutter

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The purpose of this study was to examine temperament dimensions, executive functioning ability, and anxiety levels in school-age children who stutter and their non-stuttering peers. Participants were 100 Portuguese children aged 7 to 12 years (M = 9.13; SD = 1.70), including 50 children who stutter and 50 children who do not stutter. Analyses, which were performed separately for younger and older participants, sought to identify correlations between key variables. Temperament was evaluated through a parent questionnaire, executive functioning was evaluated through children's responses on a performance test, and anxiety level was assessed through a self-perception scale. On the temperament measure, comparisons between children who stutter and their non-stuttering peers revealed that older children who stutter exhibited significantly higher scores on the Anger/Frustration, Impulsivity, and Sadness subscales, and lower averages on the Attention/Focusing, Perceptual sensitivity, and Soothability/Falling reactivity subscales. On the executive functioning task, comparisons revealed that the group of younger children who stutter exhibited significantly higher average execution times than their non-stuttering peers. There were no statistically significant differences in anxiety between children who stutter and children who do not stutter, and there were no statistically significant correlations between temperament factors and measures of executive functioning. Children who stutter experienced lower ability to orient attention and greater emotional reactivity compared with their non-stuttering peers. Significant correlations were found between executive functioning and age and among the temperament factors themselves. These results, which support the need for a multidimensional view of stuttering, were interpreted in the context of the Dual Diathesis – Stressor model. Findings indicate that temperament and executive functioning abilities may contribute to the development of stuttering.

Keywords: temperament, executive functions, anxiety, stuttering, school-age children

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INTRODUCTION

Temperament

Temperament is an overarching term for a collection of traits that are assumed to be biologically determined and related to individual differences in reactivity and self-regulation (Rothbart et al., 2000; Jones et al., 2014).

Temperament can develop over time (Goldsmith et al., 1987) and be influenced by environmental interactions (Eggers et al., 2010). According to Rothbart and colleagues, "constitutional" factors are associated with genes and environment, "reactivity" is related to sensory response systems, and "self-regulation" relates to the process that can facilitate or inhibit reactivity (Rothbart et al., 2000). Thomas and Chess (1996) described nine temperament dimensions: "Activity Level," "Rhythmicity," "Approach/Withdrawal," "Adaptability," "Threshold of Responsiveness," "Intensity of Reaction," "Quality of Mood," "Distractibility," "Attention Span," and "Persistence." The authors relate temperament to the expression of a particular behavior. Children's and adults' intrinsic motivations and abilities for a specific behavior can be mediated by aspects of their temperament, such as their activity level, their adaptability, and their persistence (Goldsmith et al., 1987). Some authors have connected temperament differences in children who stutter with their susceptibility to begin, continue, or recover from stuttering (Conture, 2001; Guitar, 2014; Ambrose et al., 2015). Specifically, studies have suggested that children with a sensitive temperament may have neural vulnerabilities that cause them to be more likely to develop stuttering (Guitar, 2014).

Findings regarding temperament in children who stutter have been inconsistent. Therefore, it is not yet possible to draw firm conclusions about differences in temperament between children who stutter and their non-stuttering peers. Still, there is an increasing literature reporting a propensity for a more reactive and sensitive temperament in children who stutter (Embrechts et al., 2000; Felsenfeld et al., 2000; Karrass et al., 2006; Eggers et al., 2010; Ambrose et al., 2015), and there is indication that more reactive and sensitive children tend to respond more strongly to disruptions in speech fluency (Walden et al., 2012).

Temperamental characteristics in preschool children that have been shown to contribute to stuttering include difficulty concentrating on tasks (Embrechts et al., 2000; Anderson et al., 2003), and low frustration tolerance (Reilly et al., 2009; Eggers et al., 2010; Druker et al., 2019). According to Spaulding et al. (2008), tasks dependent on sustained selective attention may be influenced by limited processing resources and situational demands. It is also known that attentional control plays an important role in children's ability to manage and regulate their emotions (Blair and Ursach, 2011). Several studies have reported that preschool children who stutter are prone to have difficulty adapting to new objects and situations (Embrechts et al., 2000; Anderson et al., 2003; Howell et al., 2004; Schwenk et al., 2007; Reilly et al., 2009; Eggers et al., 2010; Hollister, 2015) and have a tendency toward greater negative affect (Embrechts et al., 2000; Ntourou et al., 2013) and negative mood (Howell et al., 2004). Experimental studies of the temperament of preschool children who stutter have revealed a tendency for impulsivity (Schwenk et al., 2007; Eggers et al., 2010) and for lower self-regulation, or the ability to regulate emotional behaviors (Johnson et al., 2010; Ntourou et al., 2013).

While studies of temperament in preschool children and adults who stutter have revealed notable differences compared to peer groups who do not stutter (e.g., Reilly et al., 2009; Ntourou et al., 2013; Ambrose et al., 2015; Smith and Weber, 2017), temperament studies involving school-age children are more rare (Oyler, 1996; Nicholas et al., 2015). Those that have been conducted have shown that children of this age who stutter tend to be more sensitive and withdrawn than their non-stuttering peers (Fowlie and Cooper, 1978). There is a need to further research temperament in school-age children in order to understand the changes that arise throughout a child's development. In the same way that some studies conclude that young children and adults who stutter exhibit certain temperament characteristics, it is important to determine whether these characteristics maintain or otherwise change during the school age and how they contribute to cognitive development (Singer and Fagen, 1992).

Executive Functioning

The role of EF in childhood stuttering has been a subject of increased attention in recent years (Ntourou et al., 2013; Jones et al., 2014). EF is a term used to describe a diverse set of cognitive skills needed to perform activities that require planning and monitoring of intentional behaviors that allow individuals to interact with the world in an adaptive and appropriate way (Diamond, 2013). Researchers have highlighted three basic components of EF: inhibition, the ability to suppress a prepotent response; working memory, which implies an information updating process; and shifting, the ability to shift between tasks or mental sets and is an important aspect of executive control (Miyake et al., 2000). Despite some inconsistencies in findings across studies, several studies have shown that children who stutter, especially in earlier ages, have a tendency to be less successful in maintaining attention than their typically fluent peers (Heitmann et al., 2004; Kaganovich et al., 2010; Costelloe et al., 2015; Eichorn et al., 2017). Children who stutter are also prone to be less able to select information from sensory input (Eggers et al., 2012), more likely to exhibit impulsivity (Eggers et al., 2013), and more likely to have greater concern about their performance (Eichorn et al., 2017).

There seems to be symptoms, similar to attention deficit disorders, for some children who stutter (Anderson et al., 2003; Druker et al., 2019); however, studies related to the incidence of attention deficit disorders are not conclusive and are performed with a limited sample size (Riley and Riley, 2000; Donaher and Richels, 2012). Children who stutter tend to perform less well than their peers in working memory (Anderson and Wagovich, 2010; Oyoun et al., 2010), inhibitory control (stroop-like tasks), and attentional focusing, as indicated through parenting ratings (Wolfe and Bell, 2004; Bajaj, 2007). Difficulties related to inhibitory control and attentional focusing are especially evident in studies that use parent-report questionnaires (Ofoe et al., 2018).

Cognitive processes described above are closely linked to emotional regulation (Sudikoff et al., 2015) and can influence the experience of anxiety (Craske et al., 2009).

Anxiety

Anxiety is a general term for an individual's emotional struggle that combines nervousness, fear, apprehension, and worrying (Craske et al., 2009). According to some authors

(e.g., Craig and Hancock, 1996; Craig et al., 2003; Ezrati-Vinacour and Levin, 2004; Craig and Tran, 2014), anxiety can be divided into *trait* anxiety (related to stable anxious baseline characteristics) and *state* anxiety (related to transitory conditions due to unpleasant emotional arousal with a tendency to appear when people have to cope with demanding situations). People who stutter often struggle with *state anxiety*, since anxiety will likely become a secondary effect of living with stuttering condition rather than being a static condition (Alm and Risberg, 2007; Messenger et al., 2015). Also, according to Samochiş et al. (2011), increased anxiety is a normal reaction to the physical aspects of stuttering. Nevertheless, some studies have not supported a relationship between anxiety and stuttering or have found little significant differences (e.g., Andrews and Harris, 1964; Hedge, 1972; Andrews et al., 1983; Cox et al., 1984; Peters and Hulstijn, 1984; Craig and Hancock, 1996). Currently, the occurrence of anxiety in children who stutter is still a subject of debate (Alm and Risberg, 2007; Manning and Beck, 2013; Smith et al., 2014). Even in the literature that does support the existence of anxiety in children, the age at which anxiety symptoms begin to appear has not yet been identified. Specifically, the studies linking anxiety to preschool-age have shown no differences between children who stutter and non-stuttering peers on anxiety measures and salivary cortisol levels (van der Merwe et al., 2011). Some studies have found significantly higher anxiety symptoms in school age children who stutter, ages 7 to 12 (e.g., Iverach et al., 2011), and other studies have reported the same for children from 10 and up (Davis et al., 2007; Mulcahy et al., 2008; McAllister et al., 2015; Iverach et al., 2017). Nevertheless, other studies have not found any trend toward elevated anxiety in school age children (Andrews and Harris, 1964; Craig and Hancock, 1996; Ortega and Ambrose, 2011). Some evidence suggests that the levels of anxiety tend to increase over time and can exceed normal values in adolescence and adulthood (Mulcahy et al., 2008). Still, the meaning of these findings is unclear and according to Messenger et al. (2015), adolescents who stutter may try to present themselves positively to hide their true concerns about stuttering. This lack of consistency suggests the existence of other variables that might affect the development of anxiety.

Temperament, EF, Anxiety, and the Dual Diathesis-Stressor Model

To date, no studies have simultaneously considered the relationship between temperament, EF, and anxiety in children who stutter, even though all of these factors are believed to affect children who stutter. Because of the relationship between anxiety, temperament, and EF (Nigg, 2000), considering these factors in concert will help to elucidate how these issues relate to the development and experience of stuttering.

There is already a large body of empirical evidence suggesting a strong concurrent relationship between temperament characteristics and executive functioning (EF) (Simonds, 2006; Sudikoff et al., 2015). According to Affrunti and Woodruff-Borden (2015) the expression of temperament may be influenced by executive functioning. Temperament also includes behavioral aspects, such as approach and withdrawal, as well as attentional processes, including orientation maintenance and executive control. Together, these abilities are the building blocks of the development of self-regulation (Rothbart and Hwang, 2002). Studies of cognitive development have shown that attention control, inhibition of inappropriate behavior, decision making, and other cognitive processes that occur in emotionally demanding contexts, are strongly supported by EF (Gupta et al., 2011).

Research has further identified temperamental characteristics and cognitive abilities as predictors of anxiety (Kefalianos et al., 2012). Environmental factors can be part of these dynamic interactions and, together with temperamental characteristic and cognitive abilities, influence how children deal with stuttering. Because temperament characteristics and EF abilities may contribute to a child's likelihood of responding to experiences in a particular way, the involvement of temperament and EF in the development of stuttering can be described in terms of the dual diathesis-stressor (DD-S) model (Walden et al., 2012). The DD-S model proposes that endogenous abilities of children who stutter interact in a dynamic way with exogenous contexts (stressors). In line with this model, temperament and EF characteristics can be seen as a diathesis that can be triggered by a stressor, transforming a predisposition to an actual emotional response in a particular situation. As applied to stuttering, the theory suggests that a child's endogenous characteristics related to temperament, anxiety, and EF, may be affected by exogenous stressors that may increase (or decrease) the frequency of stuttering. Importantly, exogenous contexts (stressors) can activate cognitive and affective processes and pushing the autonomic nervous system out of homeostasis, thereby increasing the emotional response (Walden et al., 2012). This imbalance can translate into anxiety and other signs of dysregulation (Craske et al., 2009).

The present study intended to address the literature gap on the research of temperament, EF, and anxiety jointly, comparing school-age children who stutter and non-stuttering peers. The combination of these three aspects can give us further information about the interaction between emotional and cognitive factors. Moreover, the DD-S model, which focuses the interaction between intrinsic and external factors and how they may change over time, highlights the need to concurrently consider factors such as temperament, EF, and anxiety. Taken together, these factors can provide more clues about the onset, development, and possible persistence of stuttering during childhood. A better understanding of such relationships may help clinicians better understand how stuttering affects children and this understanding may contribute to the development of more effective and personalized treatment programs.

MATERIALS AND METHODS

Participants

Participants were 100 Portuguese children, 50 children who stutter ("S" Group) and 50 age-matched children who do not

stutter ("N" Group), ages 7 to 12 years old. The Stuttering Severity Instrument – 4th Edition (SSI-4) (Riley, 2009) was used to confirm and diagnose stuttering.

Table 1 shows the demographic characteristics of the participants. The sex ratio of participants who stutter was 2.6 males to each female; for participants who do not stutter, it was 0.8 males to each female. This sex ratio for children who stutter is consistent with previous literature (Craig et al., 2002; Yairi and Ambrose, 2005).

In order to explore developmental differences, the participants who stutter (n = 50) and their non-stuttering peers (n = 50) were grouped according to age: younger children (7–9 years old; M = 7.92; SD = 0.81) and older children (10–12 years old; M = 10.95; SD = 0.82).

The cutoff age point for the two groups in this study was based on the development and important changes that take place during this period, where previously acquired learning is consolidated and new intellectual, psychological and social acquisitions arise (Blake and Pope, 2008). In addition, this age group distinction corresponds to the first two education cycles in Portugal: the first cycle includes the first 4 years of school (about 7–9 years old) and the second cycle includes the 5th and 6th grades (about 10–12 years old). Depending upon a child's birth date, however, it is possible to find children in the 7th grade who are 12 years old. Pre-school education in Portugal is intended for children between 3 and 6 years old; from the age of 13, Portuguese children are usually in high school (Alarcão et al., 2009).

Inclusion and exclusion criteria ensured that children did not exhibit any neurological or psychiatric impairment, learning disorder, or history of head injury or seizures. The sample was chosen by convenience: participants who stutter were recruited from speech-language therapists and through referral of school teachers; participants who do not stutter were recruited in some schools attended by their stuttering peers. All children were monolingual speakers of Portuguese.

TABLE 1 | Demographic characteristics of the participants (children who stutter = 50; children who do not stutter = 50).

Group	Children who stutter	Children who do not stutter	Total
Age mean (SD)	9.10 (1.73)	9.16 (1.68)	9.13 (1.70)
Sex (M/F)	36/14	22/28	58/42
	(72%/28%)	(44%/56%)	(58%/42%)
Education level (n)			
1st grade	8 (16%)	3 (6%)	11 (11%)
2nd grade	11 (22%)	10 (20%)	21 (21%)
3rd grade	9 (18%)	8 (16%)	17 (17%)
4th grade	7 (14%)	14 (28%)	21 (21%)
5th to 7th grade	15 (30%)	15 (30%)	30 (30%)
Treatment (n)			
Without treatment	14 (28%)	–	–
Speech therapy	11 (22%)	–	–
Waiting or initiating	14 (28%)	–	–
Previous therapy	11 (22%)	–	–

When the study was performed, 22% of the children who stutter were in speech therapy, 22% had previous speech therapy, and 28% were waiting for therapy or just initiating speech therapy. The children who were in therapy at the time of data collection had been in treatment between 1 to 96 months (M = 9.30 mos.; SD = 19.38 mos.). Children who had previous therapy had received between 3 and 48 months of treatment (M = 13.28 mos.; SD = 12.99 mos.).

Materials

The SSI-4 (Riley, 2009) was used along with the Portuguese story, "*A história do rato Artur*" (Guimarães, 2007), "Rato Artur" story has been used in several Portuguese studies (e.g., Guimarães and Abberton, 2005; Silvestre, 2009; Silvestre et al., 2011) because it has a high test–retest consistency and is phonetically balanced. This has being interpreted that is close to spontaneous discourse (Moon et al., 2012). Eight of the 7-year-old participants had difficulties reading the story, so only the SSI-4 plates were used for those participants.

The parents provided information about socio-demographic background, and the child's stuttering via a checklist created for this study. **Table 1** shows information about children; **Table 2** shows information about parents' sex, age, education level, and family history of stuttering.

Temperament

The Temperament in Middle Childhood Questionnaire (TMCQ) (Simonds and Rothbart, 2004) is a parent reported, paper and pencil measure that evaluates temperament in middle childhood (7–10 years old). It consists of 157 questions that examine 17 dimensions of temperament: (1) Activity Level, (2) Affiliation, (3) Anger/Frustration, (4) Assertiveness/Dominance, (5) Attention Focusing, (6) Discomfort; (7) Fantasy/Openness, (8) Fear, (9) High Intensity Pleasure, (10) Impulsivity, (11) Inhibitory Control, (12) Low Intensity Pleasure, (13) Perceptual Sensitivity, (14) Sadness, (15) Shyness, (16) Soothability/Falling reactivity, (17) Activation Control (see **Table 3**). Answers are obtained by parents rating their children on a five-point Likert scale ranging from "Almost always untrue" to "Almost always true," with the option of "Does not apply."

Through the TMCQ, it is possible to identify reactivity/sensitivity and self-regulation characteristics. For example, the TMCQ scales such as Anger/frustration are connected to reactivity, whereas scales such as Inhibitory control are more related to self-regulation (Eggers et al., 2013). For example, young children may become angry and impulsive when their goals are hindered. This might occur when they have to wait for something they want (Rothbart et al., 2001).

Of the 17 dimensions of temperament that are part of the instrument, 13 dimensions derive from the well-validated Children's Behavior Questionnaire (CBQ: Rothbart et al., 2001), which has been used in several studies to investigate the relationship between temperament and stuttering (e.g., Eggers et al., 2010; Ambrose et al., 2015). In Simonds (2006), the TMCQ was shown to have good internal consistency reliability (Cronbach's alpha ranged from 0.62 to 0.83) and acceptable

TABLE 2 | Demographic characteristics of parents (parents of children who stutter = 50; parents of children who do not stutter = 50).

Group	Parents of children who stutter		Parents of children who do not stutter		Total	
Age mean (SD)	42.26 (4.82)		39.60 (4.34)		40.93 (4.76)	
Sex (M/F)	6/44 (6%/88%)		3/47 (6%/94%)		9/91/ (9%/91%)	
Family history of stuttering (n)						
Yes	30 (60%)		–		–	
No	20 (40%)		–		–	
Education level (n)	**Mother**	**Father**	**Mother**	**Father**	**Mother**	**Father**
1–4 years	0 (0%)	0 (0%)	0 (0%)	2 (4%)	0 (0%)	2 (2%)
5–6 years	0 (0%)	0 (0%)	2 (4%)	0 (0%)	2 (2%)	0 (0%)
7–9 years	5 (10%)	6 (12%)	2 (4%)	7 (14%)	7 (7%)	13 (13%)
10–12 years	8 (16%)	10 (20%)	15 (30%)	19 (38%)	23 (23%)	29 (29%)
Graduation	32 (64%)	30 (60%)	27 (54%)	21 (42%)	59 (59%)	51 (51%)
Master	3 (6%)	4 (8%)	3 (6%)	0 (0%)	6 (6%)	4 (4%)
Ph.D.	2 (4%)	0 (0%)	1 (2%)	1 (2%)	3 (3%)	1 (1%)

agreement between self-report and parent report (Pearson's r ranged from −0.02 to 0.50).

The questionnaire was translated to European Portuguese for this study (Rocha and Rato, 2017).

Executive Functioning

Children were assessed using the Portuguese version of the Children's Color Trails Test (CCTT), a neuropsychological paper and pencil test of EF (Pinto, 2008). The CCTT measures sustained visual attention, sequencing, psychomotor speed, and cognitive flexibility. It is intended for ages 8 to 16, though the authors have reported success with children as young as 7 years old (Llorente et al., 2003). The test includes two parts (CCTT-1 and CCTT-2), each involving one trial and one experimental task. In CCTT1, the child must connect the numbers from 1 to 25 following a correct sequence as quickly as possible. In CCTT2, the child must repeat the task from CCTT1 but with a color alternation. In this task, the child still connects the numbers from 1 to 25. This time, however, each number is repeated in different colors (i.e., there are yellow numbers and pink numbers), and the child must be sure to follow the numerical order even when it changes between yellow and pink (Llorente et al., 2003).

The results of both parts of this test consist of: (a) time (in seconds) that the child takes to complete the tasks, (b) the number of times almost failed (the failures), (c) the number of errors, and (d) the number of warnings (when a child makes a mistake, the examiner advises him or her to start the test again from the last correct circle).

CCTT has been increasingly used around the world (e.g., Koo and Min, 2008; Pinto, 2008; Llorente et al., 2009; Konstantopoulos et al., 2015) for the assessment of children in neurological and psychiatric disorders such as language disabilities (e.g., Williams et al., 1995), attention deficit/hyperactivity disorder (e.g., Kennel et al., 2010; Cho et al., 2011), and other conditions (Llorente et al., 2003). CCTT is based on the Trail Making Test, which assess speeded visuomotor tracking. Research has shown discriminant validity and sensitivity across cultures (Williams et al., 1995). The CCTT is expected to have the same validity as the Trail Making in the assessment of children with several disorders (Williams et al., 1995). In a study with 70 children diagnosed with attention deficit and hyperactivity disorder, CCTT exhibited appropriate test–retest reliability (Llorente et al., 2009).

Anxiety

The children also completed the Portuguese version of Multidimensional Anxiety Scale for Children (MASC), which examines the symptoms of anxiety in children and adolescents ages 7 to 19 years. It contains 39 questions, with four-point Likert scale responses (March et al., 1997; Matos et al., 2012; Salvador et al., 2017). Items on this questionnaire are grouped into four factors: (a) Physical symptoms, (b) Social anxiety, (c) Separation anxiety, and (d) Harm avoidance (Wei et al., 2014). Participants are asked to score statements such as: "I get nervous if I have to do something in public," choosing between: (a) "it is never or almost never true," (b) "it is rarely true," (c) "sometimes it is true," and (d) "It is often true."

The normative data for the MASC show that it is oriented mainly toward inherent characteristics (*trait* anxiety), though it is also influenced by transitory conditions and situations (*state* anxiety) (March et al., 1997). Decades of research confirm the robust features of the MASC. Several studies with general populations and with clinical populations supported their internal consistency, temporal stability, and convergent validity (Salvador et al., 2017). The original English version demonstrated good internal consistency (between 0.60 and 0.90), strong convergent/divergent validity, and test–retest reliability (March et al., 1997). The Portuguese version of the MASC has also been shown to be an adequate and reliable measure for self-assessment of anxious symptomatology, presenting reasonable psychometric characteristics in internal consistency, temporal stability, and validity (Salvador et al., 2017).

TABLE 3 | TMCQ scale (Simonds and Rothbart, 2004) descriptions and sample items.

TMCQ scale	Definition
Activity level	Level of gross motor activity including rate and extent of locomotion.
Affiliation	The desire for warmth and closeness with others, independent of shyness or extraversion.
Anger/ frustration	Amount of negative affect related to interruption of ongoing tasks or goal blocking.
Assertiveness/ dominance	Tendency to speak without hesitation and to gain and maintain control of social situations
Attentional focusing	Tendency to maintain attentional focus upon task-related channels.
Discomfort	Amount of negative affect related to sensory qualities of stimulation, including intensity, rate or complexity of light, movement, sound, and texture.
Fantasy/ openness	Active imagination, aesthetic sensitivity, intellectual curiosity.
Fear	Amount of negative affect, including unease, worry or nervousness related to anticipated pain or distress and/or potentially threatening situations.
High intensity pleasure	Amount of pleasure or enjoyment related to situations involving high stimulus intensity, rate, complexity, novelty, and incongruity.
Impulsivity	Speed of response initiation.
Inhibitory control	The capacity to plan and to suppress inappropriate approach responses under instructions or in novel or uncertain situations.
Low intensity pleasure	Amount of pleasure or enjoyment related to situations involving low stimulus intensity, rate, complexity, novelty, and incongruity
Perceptual sensitivity	Amount of detection of slight, low intensity stimuli from the external environment.
Sadness	Amount of negative affect and lowered mood and energy related to exposure to suffering, disappointment, and object loss.
Shyness	Slow or inhibited approach in situations involving novelty or uncertainty.
Soothability/ falling reactivity	Rate of recovery from peak distress, excitement, or general arousal.
Activation control	The capacity to perform an action when there is a strong tendency to avoid it.

Procedures

This study received full approval by the Ethics Committee of the Institute of Health Sciences of Universidade Católica Portuguesa (register number 34/2017). Prior to their participation in this study, parents signed a written informed consent for themselves and their children. Consent also included permission for the researcher to record the child and the right for participants to withdraw from the study at any time was clarified.

Children were assessed while parents completed the questionnaires. This was carried out in two sessions of approximately 30 min each.

All testing was conducted between December 2017 and May 2018. The SSI, MASC, and CCTT instruments were applied on different days and in a different order, to reduce potential order effects that might bias results.

Temperament

Temperament was assessed using the Portuguese version of the TMCQ, with the 157 original questions, distributed in 17 temperament dimensions (Simonds and Rothbart, 2004). After a brief explanation from the researcher, parents completed the TMCQ. This required approximately 20 min. In addition to researcher's explanation, on the first page of the questionnaire parents could read instructions about the content of the questions and how to complete the form. After parents completed the questionnaire, the researcher scored the instrument according to the instructions.

Executive Functioning

For the EF assessment, the researcher presented and explained to the children how to perform the CCTT1, using the trial test. In both trial test and experimental test, children drew a line between the circles following a numerical order, as fast as they could; however, the CCTT1 trial test was performed with just 8 numbers. For the CCTT2 the procedures were similar, with the difference that children should switched between colors (after a yellow circle the child should drew a line toward a pink circle, following a numerical order). The researcher recorded 9 scores for each child. These scores corresponded to: the time that the child took to complete the tests for both CCTT1 and CCTT2, as well as the number of warnings, failures, and wrong answers (Number Sequencing and Color Sequencing) (Llorente et al., 2003).

Anxiety

For the anxiety assessment, the MASC questionnaire was presented to each child. Children were asked to read all the questions and to choose the best option for each. Children were informed about the importance of responding to all questions. For 7-years-old children, the MASC questions were read in full by the examiner.

After the children completed the questionnaire, the researcher summed the items for each factor, obtaining four final scores, corresponding to: (a) Physical symptoms, (b) Social anxiety, (c) Separation anxiety, and (d) Harm avoidance, for each child.

Data Analysis

Preliminary analyses were made in order to check the assumptions of homogeneity. Results for some variables were not normally distributed; however, with the $n = 50$ for each participant group, the central limit theorem suggests that parametric tests (t-test) would still be sufficiently robust to avoid deviations from normality. Two-sample t-tests were used to compare mean scores for the stuttering and non-stuttering groups for the temperament (TMCQ), EF (CCTT), and Anxiety (MASC) measures. These analyses were performed separately for younger and older participants. A multivariate analysis using principal component analysis (PCA) was performed in order to determine which variables were correlated and to summarize children characteristics in an ordination diagram. For the PCA analyses, younger and older children were grouped. This was done because of apparent differences between age groups. The use of PCA provided a dynamic view of the interaction among all of the variables, including age. To account for the large number of variables in the study (temperament, EF, anxiety, and age) only the variables that showed statistical significance in the t-tests

were used in the PCA. Data analysis was completed using SPSS (Statistical Package for the Social Sciences – Version 24 for windows, IBM, Corp., Armonk, NY, United States).

RESULTS

Younger Children Group

Temperament

No statistically significant differences were found between groups of children who stutter and their non-stuttering peers ($p > 0.05$) for any of the variables of temperament including: (1) Activity Level, (2) Affiliation, (3) Anger/Frustration, (4) Assertiveness/Dominance, (5) Attention Focusing, (6) Discomfort; (7) Fantasy/Openness, (8) Fear, (9) High Intensity Pleasure, (10) Impulsivity, (11) Inhibitory Control, (12) Low Intensity Pleasure, (13) Perceptual Sensitivity, (14) Sadness, (15) Shyness, (16) Soothability/Falling reactivity, (17) Activation Control.

Executive Functioning

Group comparisons of the CCTT1 and the CCTT2 revealed that children who stutter exhibited significantly higher scores for execution time (CCTT1: $t_{(48.75)} = 3.144$, $p = 0.003$; CCTT2: $t_{(52.27)} = 3.753$, $p < 0.001$), as well as number of failures (CCTT1: $t_{(38.23)} = 2.627$, $p = 0.012$), number of warnings (CCTT1: $t_{(52.47)} = 2.968$, $p = 0.005$; CCTT2: $t_{(53.71)} = 3.757$, $p < 0.001$), number of sequencing errors (CCTT2: $t_{(34.99)} = 3.337$, $p = 0.002$), and color sequencing errors (CCTT2: $t_{(49.31)} = 2.416$, $p = 0.020$) (**Table 4**).

Anxiety

No statistically significant differences were found between groups of children who stutter and their non-stuttering peers ($p > 0.05$) for any of the variables of anxiety including: (1) Physical symptoms, (2) Social anxiety, (3) Separation anxiety, and (4) Harm avoidance, for each child.

Older Children Group

Temperament

Statistically significant differences were found for several temperament factors (**Table 5**). Children who stutter scored lower than non-stuttering peers in Attention/Focusing ($t_{(36)} = -3.526$, $p = 0.001$), Perceptual Sensitivity ($t_{(36)} = -2.411$, $p = 0.021$), and Soothability/Falling reactivity ($t_{(36)} = -2.932$, $p = 0.006$). Children who stutter scored higher than non-stuttering peers in temperament factors of Anger/Frustration ($t_{(36)} = 2.801$, $p = 0.008$), Impulsivity ($t_{(36)} = 2.899$, $p = 0.006$), and Sadness ($t_{(36)} = 3.683$, $p = 0.001$).

Executive Functioning

No statistically significant differences were found between groups of children who stutter and their non-stuttering peers ($p > 0.05$) for any of the variables of EF, including: (1) CCTT1 execution time, (2) CCTT1 number of sequencing errors, (3) CCTT1 number of failures, (4) CCTT1 number of warnings, (5) CCTT2 execution time, (6) CCTT2 number of color sequencing errors, (7) CCTT2 number of sequencing errors, (8) CCTT2 number of failures, (9) CCTT2 number of warnings.

TABLE 4 | Mean (M), standard deviations (SD) and *p*-values for the temperament, EF and anxiety performance tasks for group of younger children who stutter ($n = 31$; sex: M = 25; F = 6) and who do not stutter ($n = 31$; sex: M = 15; F = 16).

	Children who stutter		Children who do not stutter			
Scores	*M*	*SD*	*M*	*SD*	*t*	*p*
Activation control	3.185	0.442	3.326	0.493	−1.183	0.242
Activity level	3.632	0.755	3.794	0.709	−0.087	0.386
Affiliation	4.042	0.361	4.033	0.488	0.086	0.932
Anger/frustration	3.251	0.742	3.137	0.552	0.687	0.994
Assertiveness/ dominance	3.122	0.646	3.300	0.597	−1.124	0.265
Attention/focusing	2.840	0.993	3.513	1.990	−1.686	0.097
Discomfort	2.819	0.669	2.481	0.669	1.993	0.051
Fantasy/openness	3.766	0.660	3.857	0.552	0.586	0.560
Fear	2.804	0.689	2.612	0.606	1.167	0.298
High intensity pleasure	3.058	0.651	2.998	0.624	0.373	0.711
Impulsivity	2.983	0.544	2.959	0.523	0.184	0.854
Inhibitory control	2.962	0.575	3.110	0.587	−1.000	0.322
Low intensity pleasure	3.256	0.655	3.477	0.629	−1.359	0.179
Perceptual sensitivity	3.091	0.835	3.206	0.692	−0.591	0.557
Sadness	2.700	0.452	2.713	0.586	−0.095	0.925
Shyness	2.792	0.811	2.651	0.852	0.664	0.509
Soothability/falling reaction	3.223	0.721	3.367	0.571	−0.831	0.410
CCTT1 time (sec)	86.308	33.943	64.032	20.107	3.144	0.003**
CCTT1 number sequencing Errors	0.193	0.543	0.069	0.359	1.068	0.290
CCTT1 failures	0.548	0.961	0.065	0.359	2.627	0.012*
CCTT1 warnings	1.677	1.558	0.677	1.045	2.968	0.005**
CCTT2 time (sec)	161.420	46.582	123.677	31.061	3.753	< 0.001***
CCTT2 color sequencing errors	1.355	1.279	0.709	0.772	2.406	0.020*
CCTT2 number sequencing Errors	0.419	0.620	0.032	0.120	3.337	0.002**
CCTT2 failures	1.452	1.480	0.810	1.167	1.906	0.061
CCTT2 warnings	2.710	2.036	1.032	1.426	3.757	< 0.001***
Physical symptoms	6.258	4.885	7.267	5.836	0.733	0.467
Social anxiety	10.710	8.038	9.833	5.522	0.498	0.621
Separation anxiety	9.000	4.219	9.500	4.276	−0.460	0.647
Harm avoidance	19.774	4.566	19.700	4.276	0.065	0.948
Total score anxiety	45.420	15.000	46.267	14.694	−2.223	0.824

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.

Anxiety

As in the younger group, no statistically significant differences were found between groups of children who stutter and their non-stuttering peers ($p > 0.05$) for any of the variables of anxiety, including: (1) Physical symptoms, (2) Social anxiety, (3) Separation anxiety, and (4) Harm avoidance.

TABLE 5 | Mean (M), standard deviations (SD) and *p*-values for the temperament, EF and anxiety performance tasks for group of older children who stutter (*n* = 19; sex: M = 11; F = 8) and children who do not stutter (*n* = 19; sex: M = 7; F = 12).

	Children who stutter		Children who do not stutter			
Scores	***M***	***SD***	***M***	***SD***	***t***	***p***
Activation control	3.049	0.467	3.221	0.406	−1.205	0.236
Activity level	3.872	0.694	3.806	0.800	0.271	0.788
Affiliation	4.126	0.449	4.171	0.412	0.320	0.751
Anger/ frustration	3.335	0.614	2.807	0.546	2.801	0.008**
Assertiveness/ dominance	3.243	0.736	3.324	0.566	−3.81	0.071
Attention/ focusing	2.644	0.644	3.552	0.920	−3.526	0.001***
Discomfort	2.779	0.627	2.584	0.553	1.015	0.317
Fantasy/ openness	2.916	0.814	3.840	0.536	−1.269	0.212
Fear	2.916	0.814	2.700	6.690	0.881	0.384
High intensity pleasure	3.084	0.576	2.783	0.655	1.501	0.142
Impulsivity	3.084	0.446	2.560	0.650	2.899	0.006**
Inhibitory control	3.317	0.448	3.536	0.574	−1.312	0.198
Low intensity pleasure	3.264	0.476	3.435	0.572	−0.997	0.325
Perceptual sensitivity	3.307	0.567	3.722	0.491	−2.411	0.021*
Sadness	3.036	0.553	2.415	0.485	3.683	0.001***
Shyness	3.042	0.986	2.838	0.858	0.679	0.501
Soothability/ falling reactivity	3.157	0.402	3.663	0.636	−2.932	0.006**
CCTT1 time	51.745	15.000	52.790	19.472	−0.185	0.854
CCTT1 number sequencing errors	0.263	0.561	0.211	0.535	0.296	0.769
CCTT1 failures	0.158	0.375	0.000	0.000	1.837	0.074
CCTT1 warnings	0.632	1.065	0.211	0.419	1.604	0.118
CCTT2 times	97.9474	32.732	98.947	33.311	−0.093	0.926
CCTT2 color sequencing errors	0.579	1.610	0.421	0.838	0.379	0.707
CCTT2 number sequencing errors	0.000	0.000	0.000	0.000	0.073	0.943
CCTT2 failures	1.105	1.370	0.579	0.837	1.429	0.162
CCTT2 warnings	0.421	0.961	0.368	0.831	0.181	0.858
Physical symptoms	8.842	7.654	6.800	3.721	1.051	0.303
Social anxiety	10.263	7.001	11.526	4.937	0.642	0.525
Separation anxiety	9.474	6.040	8.526	4.033	0.569	0.573
Harm avoidance	17.947	4.972	18.158	4.375	−10.139	0.891
Total score anxiety	46.579	19.585	44.158	10.569	0.474	0.638

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.

Multivariate Analysis

The PCA ordination biplot (**Figure 1**) showed that CCTT2 Time (component loading = 0.82), CCTT2 warnings (component loading = 0.80), CCTT1 time (component loading = 0.75), age (component loading = −0.69), CCTT2 number of sequencing errors (component loading = 0.67), CCTT1 warnings (component loading = 0.65), and CCTT2 Color sequencing errors (component loading = 0.51), were the variables influencing the children's ordination along the first axis (Dimension 1), that is, the EF dimensions (**Figure 1**).

The right side of the axis shows the children with higher values of CCTT2 Time, CCTT2 warnings, CCTT1 time, CCTT2 number of sequencing errors, CCTT1 warnings, CCTT2 Color sequencing errors, and younger children. The left side of the axis shows children characterized by lower values of CCTT2 Time, CCTT2 warnings, CCTT1 time, CCTT2 number of sequencing errors, CCTT1 warnings, CCTT2 Color sequencing errors, and older children. Most of the children who stutter ("S") were plotted on the right side of the first dimension. The first axis accounted for 27.30% of the total variance. The parameters with greater contribution to the second axis (dimension 2 – Temperament dimensions) were Sadness (component loading = 0.78), Anger/Frustration (component loading = 0.75), Soothability/Falling reactivity (component loading = −0.72), Attention/Focusing (component loading = −0.69), and Impulsivity (component loading = 0.60). Most of the children who stutter were displayed on the upper part of the diagram, as they exhibited higher values of Sadness, Anger/Frustration and Impulsivity, and lower values of Soothability/Falling Reactivity and Attention/Focusing. The bottom part of the diagram shows mainly children who do not stutter, due to lower values of Sadness, Anger/Frustration and Impulsivity; and higher values of Soothability/Falling Reactivity and Attention/Focusing.

The second axis accounted for 20.04% of the total variance. The CCTT1 Time, CCTT1 Warnings, CCTT1 failures, CCTT2 Time, CCTT2 Warnings, CCTT2 number of sequencing errors, and CCTT2 color sequencing errors were highly and positively correlated with one another and negatively correlated with age. Sadness, Anger/Frustration, and Impulsivity were highly and positively correlated with each other; Attention, Soothability/Falling Reactivity and Perceptual Sensitivity were negatively correlated with Anger/Frustration, Impulsivity, and Sadness.

DISCUSSION

This study investigated temperament dimensions, EF skills, and anxiety levels in children who stutter and their non-stuttering peers. The main results are consistent with the hypothesis that some children who stutter may differ in temperament and EF factors when compared to children do not stutter. Specifically, in these group comparisons, children who stutter were found to be more reactive and sensitive than their non-stuttering peers. However, the findings were different across the two age groups that were analyzed. The differences in temperament level were

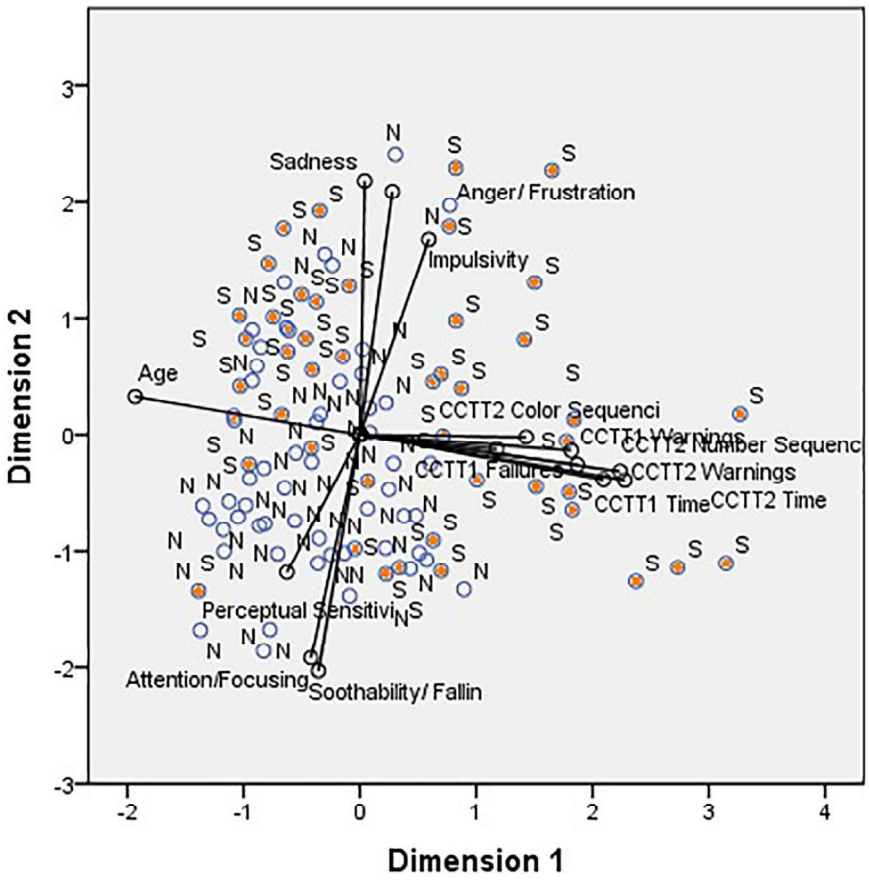

FIGURE 1 | Principal component analysis performed on children from group S and group N. Cumulative percentage variance explained By Axes: I – 27.30%; I + II – 47.34%. Groups: S – Children who stutter; N – non-stuttering children. Variables: CCTT1 Time, CCTT1 failures, CCTT1 Warnings, CCTT2 Times, CCTT2 Warnings, CCTT2 number of sequencing errors, CCTT2 color sequencing errors, Anger/Frustration, Impulsivity, Sadness, Perceptual Sensitivity, Attention/Focusing, Soothability/Falling Reactivity, and age.

noted in the group of older children only, while differences in EF were noted in the group of younger children only. Furthermore, results did not support the idea that children who stutter exhibit higher rates of anxiety than children who do not stutter, regardless of age group. Correlation analyses highlighted the dynamic nature of stuttering and suggested a link between endogenous abilities and external factors (Wolfe and Bell, 2004; Sudikoff et al., 2015).

Temperament

Results on the temperament scale are consistent with previous studies that have suggested difficulties in children who stutter compared to non-stuttering peers in attention span (Embrechts et al., 2000; Anderson et al., 2003; Eggers et al., 2010; Costelloe et al., 2015; Hollister, 2015) and a tendency toward impulsivity (Schwenk et al., 2007; Eggers et al., 2013). Attention and impulsiveness suggested a link to emotion regulation (Rothbart et al., 2001), because negative levels suggest emotional instability (Derryberry and Rothbart, 1988; Eisenberg et al., 1993). We also found differences in Anger/Frustration, Sadness, and Soothability/Falling reactivity temperament dimensions. This supports studies that indicate a more sensitive temperament in children who stutter. This could mean that school-age children who stutter may have more difficulty regulating their emotions. Furthermore, sadness could be connected to a more negative mood for children who stutter (Howell et al., 2004). A reactive temperament in children who stutter was also found in studies with preschoolers (Johnson et al., 2010; Ntourou et al., 2013) and school age children (Fowlie and Cooper, 1978). Higher scores in Anger/Frustration and lower scores in Soothability/Falling reactivity could indicate that older children who stutter (ages 10–12 years) can have more difficulty in recovering from peak distress, excitement, or general arousal (i.e., they may have a harder time settling down after an exciting activity) (e.g., Karrass et al., 2006).

Executive Functioning

Younger children who stutter required longer execution times and had a higher number of warnings and failures, number sequencing errors, and color sequencing errors compared age-matched peers who do not stutter. This suggests that children who stutter in the first years of schooling might have a lower

attention span than their peers (Anderson et al., 2003). They might also need more time to adapt to a task and to start performing (Eggers et al., 2013; Manning and Beck, 2013) or have a greater concern about errors (Eichorn et al., 2017). A higher number of failures (times when a child almost makes a mistake) may be related to the tendency for impulsivity or difficulties with inhibitory control, as has been previously suggested by some authors (Schwenk et al., 2007; Eggers et al., 2013; Ofoe et al., 2018). This was especially true for the task requiring the alternation of colors in the sequence of numbers.

Anxiety

No significant differences were detected between children who stutter and children who do not stutter in anxiety levels for either age group. According to previous studies, anxiety tends to increase as children grow older, especially between 8 to 12 years old (Blood and Blood, 2007; Messenger et al., 2015). These results are in agreement with prior researchers who reported no elevated anxiety in children who stutter (Mulcahy et al., 2008; Ortega and Ambrose, 2011; Smith et al., 2017). It could be that the participants in this study as a group showed no differences in anxiety because 22% were in speech therapy and another 22% had previously received treatment. Prior research has shown that people who are in or who have completed treatment often show comparable anxiety levels to their non-stuttering peers (Davis et al., 2002). Thus, balanced results between groups could be a consequence of the treatment itself. Other explanations may be due to methodological limitations, such as the lack of specificity of the measure to identify anxiety in the targeted population. As we saw above, anxiety in stuttering may be related to very specific situations, so, the use of a *trait* anxiety measure could have influenced the results. Speech tasks can trigger anxiety, so future research may benefit from using speech tasks rather than questionnaires (Manning and Beck, 2013; Gawda and Szepietowska, 2016). Finally, in self-report measures, children may try to give their answers a better view of themselves, trying to hide some perceived weaknesses and thereby under-reporting anxiety (Messenger et al., 2015).

Temperament, Executive Functioning, and Anxiety Interaction

Looking closely at the differences between groups, it was possible to observe different results in the older participants through the parent-perception scale and the younger participants through the performance of the EF task. It is hypothesized that Attention/Focusing, Perceptual Sensitivity, and Impulsivity issues may be subtle and unnoticed by the parents of the youngest children. Such differences may only be identifiable using sophisticated assessments such as the CCTT. In fact, some researchers agree that it is possible to find different results from behavioral measures (e.g., in novel events) and from parent reports of daily observations (Karrass et al., 2006). Moreover, parent perspectives may not reflect children's true abilities (Bernstein Ratner and Silverman, 2000), because their responses may be influenced by the emotional link that exists with children (Seifer et al., 2004). Parents may also find it easier to identify temperament characteristics as children grow older, leading to more detailed or accurate assessment of children's temperament in the older age group. The results should be interpreted with caution since the sample was not matched by gender, with sex differences being related to the fact that more females were found in the schools where the sample collection, of children who stutter, was carried out. Finally, many tasks with different sensory modalities can also influence the results (Ofoe et al., 2018). In the present study, EF was assessed using a visual search task, but for the temperament results, parents may be basing their responses on situations that are dependent on other stimuli.

Because temperament characteristics can change over time (Rothbart et al., 2000), the different pattern between two age groups in temperament dimensions could also be related to the experience of negative emotional reactions and difficulties in functional communication abilities over time (Yaruss and Quesal, 2004; Yaruss, 2010). Current results from questionnaires may indicate that parents' responses are affected by experiences rather than an inherent tendency. As older children become more aware of their stuttering, by experiencing it in different situations, they may experience greater impact of stuttering in their lives. This might exacerbate or emphasize certain characteristics to the parents' view. When correlating the various components of temperament, EF, and anxiety, it was found that, difficulties in Attention/Focusing and Soothability/Falling reactivity were correlated with a tendency toward greater sadness and Anger/Frustration. Results are in agreement with previous literature (Wolfe and Bell, 2004; Sudikoff et al., 2015) which suggests an association between the coordination and integration of mental processes in successful task performance with self-regulation of emotional states (Sudikoff et al., 2015).

Temperament, Executive Functioning, and Anxiety Interaction and the DD-S Model

Findings from the current study support the predictions from the DD-S Model (Walden et al., 2012), which state that cognitive and emotional regulation can be activated by exogenous contexts. According to the model, the cause of stuttering moments is dynamic and not just related to external factors; it also relates to how children cope with exogenous factors through endogenous abilities (Walden et al., 2012). Further research on this dynamic relationship may be a starting point for better understanding the development of stuttering and the production of individual instances of disfluency. The present study helps to further specify the predictions of the DD-S model by the potential contribution of temperament and EF as intrinsic sensitivities, which can be triggered and boosted by external agents to influence the emergence of disfluencies.

Future Directions

Because endogenous capacities, such as temperament and EF, can change over time, and because exogenous factors, such as demands of the environment, may be different for each person, future research should examine the interactions between temperament and the development of EF both individually and

over time. Similar studies that involve the analysis of several variables simultaneously may help to better explain the onset of anxiety in older children or other aspects of how stuttering – and reactions to stuttering – develop over time.

In future research, the use of multiple instruments would strengthen both the reliability and validity of these findings. For example, experimental methods that complement self-perception scales might allow the evaluation and analysis of child behavior in different situations. It would also be worthwhile to add inhibitory control and working memory tasks to better understand EF. These are the concepts that are encompassed in EF and have been examined independently in other studies (Wolfe and Bell, 2004; Oyoun et al., 2010; Eggers et al., 2013; Ntourou et al., 2017). The DD-S model predicts that emotional reactivity and emotion regulation influence the frequency and severity of stuttering in preschool-age children, so it would be appropriate for future research to examine these factors simultaneously.

Future studies should also employ a more balanced sample collection, with a more tight matching of groups in variables such as sex, age, and other relevant factors. Although this study involved a reasonable sample size, the participants were in different stages of treatment, and it is possible that participants' treatment histories might have affected the results. Similarly, the presence of some differences in sex ratio and age between sub-groups of children who stutter and children who so not stutter suggest that these preliminary results should be interpreted with caution.

CONCLUSION

Results highlight the potential role of emotional processes, temperament, and EF in the development of stuttering. Examining the cognitive and emotional skills of children who stutter across age groups can add further knowledge about stuttering. Ultimately, such knowledge may lead to refinements in clinical and educational practices. A principal outcome of this study is the finding that endogenous abilities in children who stutter may be different according to their age. Older participants were found to be more prone to difficulties in temperament dimensions, while younger participants exhibited predispositions for difficulties related to EF. This suggests that differences between children who stutter and children who do not stutter may be mediated by age and development. These results are in agreement with a dynamic view of the development of stuttering influenced by internal and external factors.

DATA AVAILABILITY STATEMENT

The datasets generated for this study are available on request to the corresponding author.

ETHICS STATEMENT

This study received full approval by the Ethics Committee of the Institute of Health Sciences of Universidade Católica Portuguesa (register number 34/2017). Prior to their participation in this study, parents signed a written informed consent for themselves and their children. Consent also included permission for the researcher to record the child and the right for participants to withdraw from the study at any time was clarified.

AUTHOR CONTRIBUTIONS

MR collected the sample, conducted the analysis, and wrote and edited the manuscript. JY contributed to the study design, analysis, and writing, editing, and reviewing of the manuscript. JR designed and supervised the study, and contributed to the writing and reviewing of the manuscript.

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Conflict of Interest: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Annexe 3

Stuttering Severity Instrument–4

SSI–4

Examiner Record Form

Glyndon D. Riley

Identifying Information

Name _____ Female ☐ Male ☐

Grade _____ Date of Birth _____

Date of testing _____ Age _____

School _____ Examiner _____

Preschool ☐ School Age ☐ Adult ☐ Reader ☐ Nonreader ☐

Frequency (Use Readers Table or Nonreaders Table, not both)

Readers Table

1. Reading Task		2. Speaking Task	
%SS	Task Score	%SS	Task Score
1	2	1	2
2	4	2	3
3–4	5	3	4
5–7	6	4–5	5
8–12	7	6–7	6
13–20	8	8–11	7
21 & up	9	12–21	8
		22 & up	9

Nonreaders Table

3. Speaking Task	
%SS	Task Score
1	4
2	6
3	8
4–5	10
6–7	12
8–11	14
12–21	16
22 & up	18

Frequency Score (use 1 + 2 or 3) ____

Duration

Average length of three longest stuttering events timed to the nearest 1/10th second		Scale Score
Fleeting	(.5 sec or less)	2
Half-second	(.6–.9 sec)	4
1 full second	(1.0–1.9 sec)	6
2 seconds	(2.0–2.9 sec)	8
3 seconds	(3.0–4.9 sec)	10
5 seconds	(5.0–9.9 sec)	12
10 seconds	(10.0–29.9 sec)	14
30 seconds	(30.0–59.9 sec)	16
1 minute	(60 sec or more)	18

Duration Score ____

Physical Concomitants

Evaluating Scale: 0 = none
1 = not noticeable unless looking for it
2 = barely noticeable to casual observer
3 = distracting
4 = very distracting
5 = severe and painful looking

Distracting Sounds:	Noisy breathing, whistling, sniffing, blowing, clicking sounds	0 1 2 3 4 5 ____
Facial Grimaces:	Jaw jerking, tongue protruding, lip pressing, jaw muscles tense	0 1 2 3 4 5 ____
Head Movements:	Back, forward, turning away, poor eye contact, constant looking around	0 1 2 3 4 5 ____
Movements of the Extremities:	Arm and hand movement, hands about face, torso movement, leg movements, foot-tapping, or swinging	0 1 2 3 4 5 ____

Physical Concomitants Score ____

Total Score

Frequency _____ + Duration _____ + Physical Concomitants _____ = ____ Percentile _____ Severity _____

3 4 5 6 7 8 9 10 17 16 15 14 13 12 11 10 09

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Annexe 4

A história do rato Artur

Havia uma vez um rato chamado Artur, que era muito indeciso. Sempre que os seus amigos lhe perguntavam se queria sair com eles, ele apenas respondia:

- Não sei.

Ele nunca dizia que "sim", nem que "não". Não era capaz de tomar decisões.

A sua tia Helena disse-lhe:

- Ninguém se interessará por ti se continuares a ser assim. Tu não tens mais cabeça do que uma folha de erva. Artur olhou atento mas não disse nada.

Num dia chuvoso os ratos ouviram um grande barulho no sótão onde viviam. As traves de pinho estavam todas estragadas e por fim uma das juntas cedeu e caiu ao chão. As paredes tremiam e os ratos ficaram com os pelos em pé, cheios de medo e terror.

- Isto não dá, disse o rato velho que era o chefe,

- Vou mandar escuteiros lá para fora à procura de uma nova casa.

Três horas mais tarde os sete escuteiros voltaram e disseram:

- Encontrámos uma casa de pedra que é mesmo o que nós queríamos. Há espaço e boa comida para todos nós. Há lá um cavalo, simpático, chamado Nelly, uma vaca, um novilho e um jardim com uma árvore.

De repente, o rato velho lembrou-se do jovem Artur.

- Vens connosco? perguntou.

- Não sei, suspirou Artur, "Talvez o telhado não venha abaixo já".

- Bom, disse o rato velho zangado, "Não podemos esperar todo o dia até que te decidas. Toca a andar! Marchar!" e foram-se embora.

Artur ficou e viu os outros ratos a irem embora cheios de pressa. A ideia de uma decisão imediata era de mais para ele.

- Vou voltar para o meu buraco por um momento, disse para ele mesmo, "só para me decidir".

Nessa noite houve uma grande tempestade que fez tremer a terra, e o telhado veio todo abaixo. No dia seguinte, vieram uns homens ver as ruínas. Um deles moveu uma tábua, e debaixo dela viram um rato jovem deitado de lado, morto, meio dentro e meio fora do seu buraco.

Versão Portuguesa (Guimarães, 2002) - «The story of Arthur the Rat» (Abercombrie, 1967)

Annexe 5

Sujeito:_____

MASC –J. March, 1997
Tradu´²o – A. Baptista & M. Carvalho, 1998

Este question®rio faz perguntas acerca do modo como tu tens pensado, sentido ou comportado recentemente. Em frente a cada uma das frases faz uma cruz em cima do nßmero que melhor mostra at¶ que ponto a frase ¶verdade para ti. Se a frase ¶verdade para ti muitas vezes faz a cruz em cima do nßmero 3. Se ¶verdade para ti algumas vezes faz a cruz no nßmero 2. Se ¶verdade uma vez por outra faz a cruz no nßmero 1. Se a frase quase nunca ¶verdade faz a cruz no nßmero 0. Lembra-te que n²o existem respostas certas nem erradas, responde apenas a propÆsito do modo como te tens sentido recentemente.

Tens a seguir dois exemplos para te ajudar a preencher o question®rio. No exemplo A, se raramente tens medo de c²es, faz a cruz em cima do 1, o que quer dizer que a frase raramente ¶verdade para ti. No exemplo B, se as trovoadas te perturbam algumas vezes, faz a cruz no nßmero 2, para dizer que a frase ¶ algumas vezes verdade para ti.

	Nunca ou quase nunca ¶ verdade	Raramente ¶ verdade	Algumas vezes ¶ verdade	Frequentemente ¶verdade
Exemplo A. Tenho medo de c²es……..	0	1	2	3
Exemplo B. As trovoadas perturbam-me…….	0	1	2	3

Responde agora a todas as questÞes que se seguem:

	Nunca ou quase nunca ¶ verdade	Raramente ¶ verdade	Algumas vezes ¶ verdade	Frequentemente ¶verdade
1. Sinto-me tenso ou com os mßsculos rºgidos……..	0	1	2	3
2. Pe´o habitualmente permiss²o antes de fazer alguma coisa……..	0	1	2	3
3. Preocupo-me por os outros se rirem de mim…….	0	1	2	3
4. Fico assustado quando os meus pais saem……	0	1	2	3
5. Tenho dificuldades em respirar…….	0	1	2	3
6. Mantenho-me atento ao perigo……..	0	1	2	3
7. A ideia de ir acampar assusta-me……..	0	1	2	3
8. Fico tr¶mulo ou agitado……	0	1	2	3
9. Esfor´o-me por obedecer aos meus pais e professores	0	1	2	3
10. Tenho medo que os outros jovens gozem comigo..	0	1	2	3
11. Tento estar perto da minha m²e ou do meu pai….	0	1	2	3
12. Tenho tonturas ou sensa´Þes de desmaio……	0	1	2	3
13. Verifico as coisas antes de as fazer……	0	1	2	3
14. Preocupo-me por me fazerem perguntas na aula...	0	1	2	3
15. Assusto-me com facilidade……	0	1	2	3
16. Tenho medo que os outros pensem que eu sou estßpido…….	0	1	2	3
17. Durmo com a luz acesa……	0	1	2	3

Annexe 6

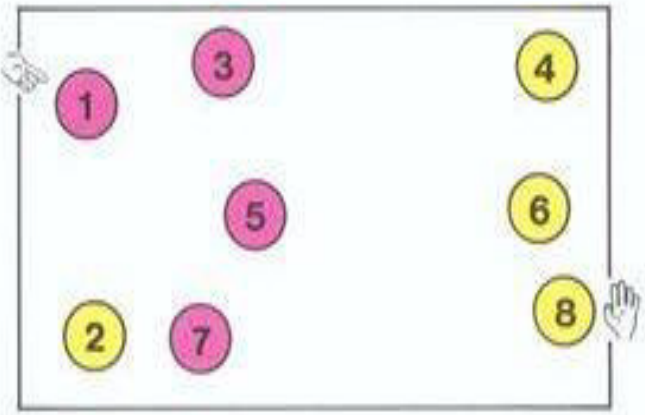
Color Trails 1

by Louis F. D'Elia, PhD, and Paul Satz, PhD

Form A

Name: _____

ID#: _____ Date: _____



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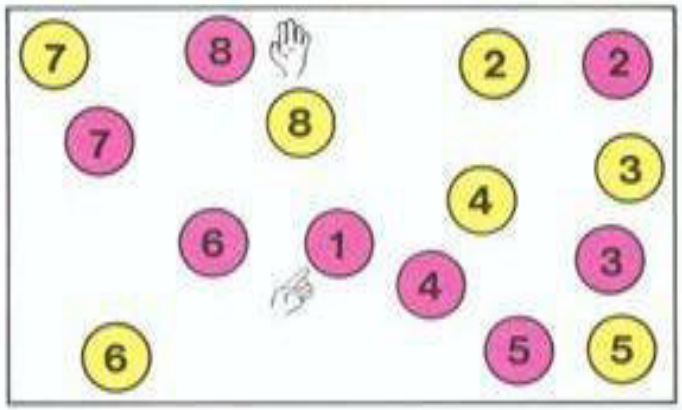
Color Trails 2

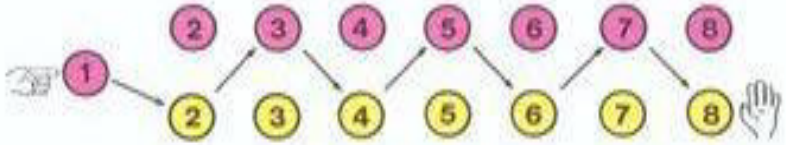
by Louis F. D'Elia, PhD, and Paul Satz, PhD

Form A

Name: _____

ID#: _____ Date: _____





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Annexe 7

Temperament in Middle Childhood Questionnaire (Version 3.0)

Today's Date _____ **Sex of Child** M F (circle one)

Child's Height _____ / _____ **Child's Date of Birth** _____
Feet / Inches Month Day Year

Staff Use: Subj. No. _____ **Age of Child** _____ Years / _____ Months

Race/Ethnicity of Child: European American/White ___ Hispanic ___

African-American ___ Asian/Pacific Islander ___ Native American/Indian ___

Multiracial ___ Other (please specify) _____

Your Relationship to Child:
Mother _____ **Father** _____
Other: (please indicate relationship**)** _____
_____

Instructions: Please read carefully before starting:

On the next pages you will see a set of statements that describe children's reactions to a number of situations. We would like you to tell us what your child's reaction is likely to be in those situations. There are of course no "correct" ways of reacting; children differ widely in their reactions, and it is these differences we are trying to learn about. Please read each statement and decide whether it is a "true" or "untrue" description of your child's reaction within the past six months. Use the following scale to indicate how well a statement describes your child:

Circle #	If the statement is:
1	Almost always untrue of your child
2	Usually untrue of your child
3	Sometimes true, sometimes untrue of your child
4	Usually true of your child
5	Almost always true of your child

If you cannot answer one of the items because you have never seen the child in that situation, for example, if the statement is about the child playing wildly and recklessly and you have never seen your child play that way, then circle NA (not applicable).

Please be sure to respond by circling a number or NA for every item. If you find an item objectionable or upsetting, you may make an exception to this instruction and skip the item.

Appendices

Appendice 1

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE
LISBOA·PORTO·VISEU

Orientações para recolha de dados

Investigação: School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life' ('crianças portuguesas em idade escolar que gaguejam – desempenho socio-cognitivo e impacto da gaguez na sua qualidade de vida'), com a orientação da Professora Doutora Joana Rato (ICS-UCP) e professor Doutor Scott Yaruss (University of Pittsburgh) tem como objetivos:

Objetivos gerais da investigação:

- Traduzir e adaptar o Overall Assessment of the Speaker's Experience of Stuttering (OASES-S) (Yaruss & Quesal, 2010) para a população Portuguesa;
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- Comparar os resultados do impacto da gaguez da perspetiva das crianças com a perspetiva dos pais/professores;
- Determinar dados normativos portugueses para o débito verbal e articulatório;
- Investigar as funções executivas, temperamento e ansiedade nas crianças portuguesas, em idades escolar, que gaguejam e comparar o seu desempenho com crianças, em idade escolar, que não gaguejam.

A Recolha de dados deverá ser realizada junto da criança (em dois momentos, ou seja, implica duas sessões, em dias diferentes), junto do Encarregado de Educação (num único momento) e do Professor (num único momento) e deverá ser realizada, somente, após autorização dos encarregados de educação, que deverá ler e assinar o documento que corresponde ao **consentimento informado.**

Momento 1: Criança

1) Em primeiro lugar deve proceder-se à gravação do discurso da criança (mínimo 200 sílabas) com utilização das **pranchas do Stuttering Severity Instrument SSI-4** (Riley, 2009). A gravação deverá ser preferencialmente vídeo, mas caso não seja possível deve existir uma descrição dos comportamentos secundários (caso existam). O/a avaliador/a deve mostrar as pranchas à criança e pedir para ela descrever as mesmas: "Explica-me o que vês nas imagens". O/a avaliador/a poderá realizar questões, de forma a estimular o discurso da criança. Existem alguns absurdos nas duas primeiras pranchas, pelo que o/a avaliador/a deverá dizer: "Olha, há algumas imagens que são um bocadinho tontas";
2) Em segundo lugar procede-se à gravação da leitura do texto: **A história do Rato Artur** (Guimarães, 2002) (não aplicar a crianças com 7 anos de idade, excepto se a aprendizagem da leitura e da escrita tenha ocorrido precocemente);
3) No caso de a gravação ser áudio, devem ser contabilizados e descritos, no momento, os comportamentos secundários. Seguem exemplos de comportamentos secundários: 1) sons distrativos (respiração audível, assobio, fungar, soprar, estalidos; 2) expressões faciais (movimento da mandíbula, protusão da língua, pressão dos lábios, tensão muscular da mandíbula); 3) movimentos da cabeça (para trás, para a frente, à volta, pobre contacto ocular, olhar em volta constantemente); 4) movimentos das extremidades (movimentos do braço e da mão, mão na cara, movimentos do tronco, movimentos das pernas, batimentos ou balanceamento dos pés);
4) Depois de realizadas as gravações deve seguir-se a aplicação do questionário **Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa (OASES-S-PT)** (Yaruss & Quesal, 2010). Deve pedir-se à criança para ler todas as questões e escolher a opção com que mais se identifica. A criança deverá ser informada da importância de responder a todas as questões. Caso haja alguma dúvida a criança poderá pedir ajuda ao/à avaliador/a. Para as crianças com 7 anos de idade as questões devem ser lidas na íntegra. É necessário contabilizar o tempo que a criança demorou a preencher o questionário.

Momento 2: Criança

3) Aplicar o questionário **"Escala Multidimensional de Ansiedade para Crianças" (MASC) (JMarch, 1997)** traduzido e adaptado de A. Baptista & M. Carvalho (1998). Deve pedir-se à criança para ler todas as questões e escolher a opção com que mais se identifica. A criança deverá ser informada da importância de responder a todas as questões. Caso haja alguma dúvida a criança poderá pedir ajuda ao/à avaliador/a/. As questões devem ser lidas na íntegra a crianças com 7 anos de idade.

4) Em último lugar será realizada a aplicação do *Children s Color Trails Test* (CCTT) (Llorente, Williams, Satz, & D'Elia, 2003). **Esta aplicação é realizada pela investigadora,** pelo que deverá ser agendada uma data para a aplicação do mesmo.

Momento 1: Pais

1) O/A colaborador/a não necessita de estar junto dos pais. Enquanto está com a criança, poderá entregar os questionários aos pais e pedir para que preencham os mesmos. Os questionários para os pais seguem em envelope selado. Deve pedir-se para ler todas as questões e escolher a opção que se relaciona mais com o/a seu/sua filho/a. Os pais devem ser informados da importância de responder a todas as questões. É necessário contabilizar o tempo que a pessoa demora a preencher o **questionário de caraterização sociodemográfico; questionário Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa – Adaptação para pais** (Yaruss & Quesal, 2010) e o **questionário de Temperamento na Terceira Infância** (Simonds & Rothbart, 2004) traduzido e adaptado para o Português Europeu por M. Rocha & J. Rato (2017);
2) No envelope que diz "pais" constam os seguintes documentos: a) **questionário de caraterização sociodemográfico; b) questionário Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa – Adaptação para pais** (Yaruss & Quesal, 2010) (OASES-S-PT-P).c) **questionário de Temperamento na Terceira Infância** (Simonds & Rothbart, 2004) traduzido e adaptado para o português euroeu por M. Rocha & J. Rato (2017);

Nota: A aplicação dos questionários deverá ser realizada preferencialmente e sempre que possível, com a mãe da criança, de forma a evitar que as diferenças de género possam influenciar a investigação;

Momento 1: Professores

1) O colaborador/a não necessita de estar presencialmente com os professores. Deve pedir-se aos pais para entregar o envelope que diz "professor" ao professor titular da criança. No envelope constam: **a) carta de apresentação b) questionário de caraterização sociodemográfico; c) questionário Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa – Adaptação para professores (Yaruss & Quesal, 2010) (OASES-S-PT-Prof).**

Os seguintes documentos/instrumentos seguem via correio:

1. Declaração de consentimento informado;
2. Termo de Anuência
3. Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa (OASES-S-PT);
4. Pranchas do SSI-4
5. História do Rato Artur;
6. Questionário "Escala Multidimensional de Ansiedade para Crianças (MASC)".
7. Envelope "pais": a) Questionário de Caraterização Sociodemográfico; b) Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa – Adaptação Pais (OASES-S-PT-P); c) Questionário de Temperamento na Terceira Infância
8. Envelope "professores": a) Carta de Apresentação; b) Questionário de Caraterização Sociodemográfico; b)Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa (OASES-S-PT-Prof)

Obrigada pela colaboração,

Mónica Rocha – Estudante de Doutoramento na UCP

Appendice 2

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LISBOA·PORTO·VISEU

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1) Em primeiro lugar deve aplicar-se o questionário **Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa (OASES-S-PT)** (Yaruss & Quesal, 2010). Deve pedir-se à criança para ler todas as questões e escolher a opção com que mais se identifica. A criança deverá ser informada da importância de responder a todas as questões. Caso haja alguma dúvida a criança poderá pedir ajuda ao/à avaliador/a. Para as crianças com 7 anos de idade as questões devem ser lidas na íntegra. É necessário contabilizar o tempo que a criança demorou a preencher o questionário.
2) Em seguida realizam-se as gravações áudio ou vídeo do discurso espontâneo e da leitura.
3) Deve proceder-se à gravação do discurso da criança (mínimo 200 sílabas) com utilização das **pranchas do Stuttering Severity Instrument SSI-4** (Riley, 2009). A gravação deverá ser preferencialmente vídeo, mas caso não seja possível deve existir uma descrição dos comportamentos secundários (caso existam). O/a avaliador/a deve mostrar as pranchas à criança e pedir para ela descrever as mesmas: "Explica-me o que vês nas imagens". O/a avaliador/a poderá realizar questões, de forma a estimular o discurso da criança. Existem alguns absurdos nas duas primeiras pranchas, pelo que o/a avaliador/a deverá dizer: "Olha, há algumas imagens que são um bocadinho tontas";
4) Por fim procede-se à gravação da leitura do texto: **A história do Rato Artur** (Guimarães, 2002) (não aplicar a crianças com 7 anos de idade, excepto se a aprendizagem da leitura e da escrita tenha ocorrido precocemente);
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Obrigada pela colaboração,

Mónica Rocha – Estudante de Doutoramento na UCP

Appendice 3

OASES™

Overall Assessment of the Speaker's Experience of Stuttering

Avaliação Global da Experiência Subjetiva da Gaguez

OASES-S

European Portuguese/Português Europeu
Formulário de Resposta
Idade 7-12

J. Scott Yaruss, PhD, CCC-SLP, BCS-F, F-ASHA
Craig E. Coleman, MA, CCC-SLP, BCS-F
Robert W. Quesal, PhD, CCC-SLP, BCS-F, F-ASHA

Traduzido por:
Mónica Rocha, SLP, Estudante de PhD com a colaboração das Terapeutas da Fala:
Joana Caldas, Elsa Margarido, Margarida Morgado, Maria João Morgado
Orientação de Joana Rato, PhD, UCP

Nome: _____ Sexo/Género: _____

Data de Nascimento: _____/_____/_____ Idade: _____ Ano de escolaridade: _____

Data: _____/_____/_____ Número de identificação: _____

Instruções gerais:

Este formulário inclui quatro secções de perguntas sobre a tua experiência *atual* com a tua fala e gaguez. Para cada questão, faz um círculo à volta o item que mais se adequa a ti. Por favor, responde a todas as questões. Se uma determinada questão não se aplicar a ti, faz uma cruz na caixa e passa para o item seguinte. Se não tens a certeza sobre como responder a qualquer uma das questões, deves pedir ajuda. Se preferires, um adulto poderá ler-te as questões.

Pontuação: Exclusivo para uso clínico

Instruções para clínicos:

- Calcular a Pontuação de Impacto das quatro secções do OASES-S, primeiro somando o número de pontos de cada secção (A) e depois contando o número de itens completos em cada secção (B). Dividir o número total de pontos (A) pelo número de itens completos (B) para obter a Pontuação de Impacto.
- Calcular a Pontuação de Impacto Global, somando os números da coluna (A) e (B) no final de cada coluna. Dividir a soma de (A) pela soma de (B) para obter a Pontuação de Impacto Global.
- A Pontuação de Impacto varia entre 1.0 e 5.0. Circundar a pontuação correspondente à avaliação de impacto para cada secção e para o Impacto Global.

	Pontuação de Impacto			Avaliação de Impacto				
	A Pontos	B Itens respondidos	A ÷ B = Pontuação de Impacto	Pontuação 1.00–1.49	Pontuação 1.50–2.24	Pontuação 2.25–2.99	Pontuação 3.00–3.74	Pontuação 3.75–5.00
Secção I: **Informação geral**	_____ ÷	_____ =	_____	Ligeiro	Ligeiro-moderado	Moderado	Moderado-grave	Grave
Secção II: **Reações da pessoa**	_____ ÷	_____ =	_____	Ligeiro	Ligeiro-moderado	Moderado	Moderado-grave	Grave
Secção III: **Comunicação diária**	_____ ÷	_____ =	_____	Ligeiro	Ligeiro-moderado	Moderado	Moderado-grave	Grave
Secção IV: **Qualidade de vida**	_____ ÷	_____ =	_____	Ligeiro	Ligeiro-moderado	Moderado	Moderado-grave	Grave
Impacto Global:	_____ ÷	_____ =	_____	Ligeiro	Ligeiro-moderado	Moderado	Moderado-grave	Grave

Stuttering Therapy Resources™

Appendice 4

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE

LISBOA·PORTO·VISEU

TERMO DE ANUÊNCIA

Instrumento: Overall Assessment of the Speakers's Experience of Stuttering – Ages 7-12 (OASES-S).

Autor original: Yaruss & Quesal, 2010

Tradução e adaptação para o Português Europeu por : Mónica Rocha, Doutoranda na Universidade Católica Portuguesa com orientação da Professora Joana Rato, PhD, Universidade Católica Portuguesa e J. Scott Yaruss, Phd, University of pittsburgh

Declaro que tenho conhecimento que o OASES-S é propriedade exclusiva intelectual dos autores e está interdita a sua circulação. Aceito que o seu uso seja exclusivo no âmbito do trabalho de investigação. Concordo com a cedência dos dados recolhidos de forma a estabelecer-se parceria com o projecto: «School age Portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life», em desenvolvimento pela Investigadora Mónica Rocha. Para o efeito foi-me facultado todo o material de aplicação e folhas de resposta que devo devolver com o término da colaboração.

Colaborador/a na investigação

Mónica Rocha

Mónica Rocha

Estudante de Doutoramento

Appendice 5

Sujeito:_____

Questionário de Caraterização Sociodemográfico e Clínico

(Encarregado de Educação)

O presente questionário foi elaborado especificamente no âmbito do projeto de doutoramento intitulado School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life', com a orientação da Professora Doutora Joana Rato (ICS-UCP) e Professor Doutor Scott Yaruss (University of Pittsburgh).

O presente questionário tem como objetivo recolher informações pertinentes relativas à criança que irá participar no estudo e ao encarregado de educação. O preenchimento deste questionário demorará cerca de 5 minutos. Agradecemos que responda a TODAS as questões.

Questionário de Caraterização Sociodemográfico e Clínico

Assinale com uma cruz (x) e/ou preencha os espaços das questões apresentadas

Dados – Encarregado de Educação

1. **Idade:** _____**2. Género:** Masc. ☐ Fem. ☐ 3. **Grau de parentesco**_____
4. **Profissão**: _____
5. **Nacionalidade**: _____
6. **Concelho de residência**_____
7. **Habilitações Literárias** (pai) 1º ciclo (1-4 anos) ☐ 2º ciclo (5-6anos) ☐ 3º ciclo (7-9 anos) ☐ Secundário (10-12 anos) ☐ Licenciatura ☐ Mestrado ☐ Doutoramento ☐
8. **Habilitações Literárias** (mãe) 1º ciclo (1-4 anos) ☐ 2º ciclo (5-6anos) ☐ 3º ciclo (7-9 anos) ☐ Secundário (10-12 anos) ☐ Licenciatura ☐ Mestrado ☐ Doutoramento ☐

Preencher só se Encarregado de Educação não for um dos progenitores

9. **Habilitações Literárias** (Enc. Ed.) 1º ciclo (1-4 anos) ☐ 2º ciclo (5-6anos) ☐ 3º ciclo (7-9 anos) ☐ Secundário (10-12 anos) ☐ Licenciatura ☐ Mestrado ☐ Doutoramento ☐

Dados - Educando

1. **Data de Nascimento: ___/___/___**
2. **Nacionalidade: _____3. Língua/s materna/s _____**

4. **A criança apresenta indícios/encontra-se sinalizada com:**

Alteração Neurológica ☐ **Défice Auditivo** ☐ **Défice Cognitivo** ☐

Outra ☐ **Qual? _____**

5. **Com que idade o/a seu/sua filho/a aprendeu a ler/escrever?**

_____

6. **Há quanto tempo é que a criança gagueja?**

_____

7. **Existe alguém na família que gagueje ou já gaguejou?**

Sim ☐

Não ☐

8. **A criança tem alguma consciência da sua gaguez?**

Sim ☐

Não ☐

9. **Costuma conversar com o/a seu/sua filho/a sobre a gaguez?**

Frequentemente ☐

Nunca ☐

Às vezes ☐

Raramente ☐

10. **A criança já realizou alguma intervenção para gaguez? Se sim, qual?**

Sim ☐ _____ Durante quanto tempo? _____

Não ☐

Appendice 6

Sujeito:_____

Questionário de Caraterização Sociodemográfico e Clínico

(Encarregado de Educação)

O presente questionário foi elaborado especificamente no âmbito do projeto de doutoramento intitulado School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life', com a orientação da Professora Doutora Joana Rato (ICS-UCP) e Professor Doutor Scott Yaruss (University of Pittsburgh).

O presente questionário tem como objetivo recolher informações pertinentes relativas à criança que irá participar no estudo e ao encarregado de educação. O preenchimento deste questionário demorará cerca de 5 minutos. Agradecemos que responda a TODAS as questões.

Questionário de Caraterização Sociodemográfico e Clínico

Assinale com uma cruz (x) e/ou preencha os espaços das questões apresentadas

Dados – Encarregado de Educação

1. **Idade:** _____**2. Género:** Masc. ☐ Fem. ☐ 3. **Grau de parentesco**_____
4. **Profissão**: _____
5. **Nacionalidade**: _____
6. **Concelho de residência**_____
7. **Habilitações Literárias** (pai) 1º ciclo (1-4 anos) ☐ 2º ciclo (5-6anos) ☐ 3º ciclo (7-9 anos) ☐ Secundário (10-12 anos) ☐ Licenciatura ☐ Mestrado ☐ Doutoramento ☐
8. **Habilitações Literárias** (mãe) 1º ciclo (1-4 anos) ☐ 2º ciclo (5-6anos) ☐ 3º ciclo (7-9 anos) ☐ Secundário (10-12 anos) ☐ Licenciatura ☐ Mestrado ☐ Doutoramento ☐

Preencher só se Encarregado de Educação não for um dos progenitores

9. **Habilitações Literárias** (Enc. Ed.) 1º ciclo (1-4 anos) ☐ 2º ciclo (5-6anos) ☐ 3º ciclo (7-9 anos) ☐ Secundário (10-12 anos) ☐ Licenciatura ☐ Mestrado ☐ Doutoramento ☐

Dados - Educando

1. **Data de Nascimento: ___/___/___**
2. **Nacionalidade: _____3. Língua/s materna/s _____**

4. **A criança apresenta indícios/encontra-se sinalizada com:**

Alteração Neurológica ☐ **Défice Auditivo** ☐ **Défice Cognitivo** ☐

Outra ☐ **Qual? _____**

5. **Com que idade o/a seu/sua filho/a aprendeu a ler/escrever?**

   _____

6. **O/A seu/sua filho/a gagueja?**

   _____

Appendice 7

Sujeito:_____

Questionário de Caraterização do/a Aluno/a e Professor/a

O presente questionário foi elaborado especificamente no âmbito do projeto de doutoramento intitulado 'School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life' ('Crianças portuguesas em idade escolar que gaguejam – Desempenho socio-cognitivo e impacto da gaguez na sua qualidade de vida'), com a orientação da Professora Doutora Joana Rato (ICS-UCP) e Professor Doutor Scott Yaruss (University of Pittsburgh).

O presente questionário tem como objetivo recolher informações pertinentes relativas à criança que irá participar no estudo e ao/à seu/sua professor/a. O preenchimento deste questionário demorará cerca de 5 minutos. Agradecemos que responda a TODAS as questões.

Questionário de Caraterização do/a Aluno/a e Professor/a

Assinale com uma cruz (x) e/ou preencha os espaços das questões apresentadas

1. **Há quanto tempo está na docência?**

   _____

2. **Há quanto tempo é professor/a do/a aluno/a em questão?**

   _____

3. **Este é o/a seu/sua primeiro/a aluno/a que gagueja? Se a resposta for não, por favor indique quantos/as alunos/as que gaguejam já fizeram parte das suas turmas, ao longo dos anos?**

   Sim ☐

   Não ☐ Número de alunos/as: _____

4. **Já alguma vez abordou a questão da gaguez com os pais do/a aluno/a/?**

   Apenas uma vez ☐ Frequentemente ☐ Nunca ☐ Às vezes ☐ Raramente ☐

5. **Já alguma vez abordou a questão da gaguez com o/a aluno/a?**

   Apenas uma vez ☐ Frequentemente ☐ Nunca ☐ Às vezes ☐ Raramente ☐

6. **Ter um/uma aluno/a que gagueja, interfere com o funcionamento normal da sua sala? Se a resposta for sim, por favor indique de que forma.**

   Sim ☐

   Não ☐

   _____

7. **Tem conhecimentos teóricos sobre gaguez?**

   Sim ☐

   Não ☐

   Se respondeu sim, por favor indique onde adquiriu a informação:

   Formação académica ☐ Pesquisas online ☐ Participação em workshops/formação sobre o tema ☐ Outra ☐ Por favor indique qual/quais _____

Appendice 8

Sujeito:_____

Question®rio de Temperamento na Terceira Inf°ncia

Instru´Þes: Por Favor leia cuidadosamente antes de come´ar:

Nas prÆximas p®ginas ver® um conjunto de afirma´Þes que descrevem as rea´Þes da crian´a a um nßmero de situa´Þes. Gostarºamos que nos dissesse como ¶ que a sua crian´a costuma reagir nessas situa´Þes. Obviamente, que n² o existem formas "corretas" de reagir; as crian´as diferem muito nas suas rea´Þes, e ¶ sobre essas diferen´as que nÆs estamos a tentar aprender mais. Por favor leia cada afirma´² o e decida se ¶ uma descrição "verdadeira" ou "falsa" para a reaç² o da sua crian´a, nos ßltimos seis meses. Use a escala seguinte para indicar o qu² o bem a afirma´² o descreve a sua crian´a.

Assinale #	se a afirma´² o ¶:
1	Quase sempre falsa para a sua crian´a
2	Geralmente falsa para a sua crian´a
3	ũs vezes verdadeira, outras vezes falsa para a sua crian´a
4	Geralmente verdadeira para a sua crian´a
5	Quase sempre verdadeira para a sua crian´a

Se n² o consegue responder a um dos itens porque nunca viu a crian´a nessa situa´² o, por exemplo, se a afirma´² o ¶ sobre a crian´a a brincar loucamente e imprudentemente e nunca a viu a brincar dessa forma, ent² o assinale com um cºrculo na op´² o NA (n² o aplic®vel).

Certifique-se de responder assinalando um nßmero ou a op´² o NA para cada item. Se encontrar um item question®vel ou perturbador, pode fazer uma exce´² o a esta instru´² o e saltar o item.

Appendice 9

Sujeito:_____

Avaliação Global da Experiência da Gaguez pela pessoa – Versão Portuguesa -Adaptação para pais-

Secção I: Informação Geral

Para cada item desta secção, faça um círculo à volta do número que se aplica ao/à seu/sua filho/a. Pense nas experiências *atuais* e sentimentos do/a seu/sua filho/a quando estiver a responder a cada questão. Se a questão não se aplica ao/à seu/sua filho/a, faça uma cruz na caixa e passe para a questão seguinte.

A. Informação geral sobre a fala do/a seu/sua filho/a	**Sempre**	**Muitas Vezes**	**Às vezes**	**Raramente**	**Nunca**
1. Com que frequência o/a seu/sua filho/a é capaz de falar fluentemente (sem gaguejar)?	1	2	3	4	5
2. Com que frequência a fala do/a seu/sua filho/a lhe parece "natural"? (como a fala das outras crianças)	1	2	3	4	5
3. Com que frequência o/a seu/sua filho/a usa as estratégias ou técnicas aprendidas na terapia da fala? (Se nunca teve terapia da fala, selecione "não teve/tem terapia da fala"	1	2	3 ☐ Não teve/tem terapia da fala	4	5
4. Com que frequência o/a seu/sua filho/a diz *exatamente* o que quer dizer, mesmo que pense que pode gaguejar?	1	2	3	4	5

B. Na sua perspetiva, quanto o/a seu/sua filho/a *sabe* sobre…?	**Imenso**		**Alguma coisa**		**Nada**
5. gaguez em geral	1	2	3	4	5
6. o que ajuda as pessoas a gaguejar *menos*	1	2	3	4	5
7. o que faz as pessoas gaguejar *mais*	1	2	3	4	5
8. o que acontece à sua fala quando gagueja	1	2	3	4	5
9. grupos de apoio ou de auto-ajuda para crianças se juntarem e falarem sobre gaguez (grupos de miúdos que se juntam e falam sobre gaguez)	1	2	3	4	5

C. De forma geral, na sua perspetiva, como o/a seu/sua filho/a se sente em relação…?	**Muito Bem**	**Bem**	**Nem bem, nem mal**	**Mal**	**Muito Mal**
10. à sua fala, em geral	1	2	3	4	5
11. a falar com as pessoas, em geral	1	2	3	4	5
12. às técnicas ou ferramentas que aprendeu na terapia da fala (Se nunca teve terapia da fala, selecione "não teve/tem terapia da fala")	1	2	3 ☐ Não teve/tem terapia da fala	4	5
13. a ser uma criança que gagueja	1	2	3	4	5
14. a ser chamado/a de gago/a pelas outras pessoas	1	2	3	4	5
15. a grupos de apoio ou de auto-ajuda para crianças que gaguejam	1	2	3	4	5

OASES-S – Overall Assessment of the speaker's Experience of Stuttering, Yaruss, S. & Quesal, R. (2010). Questionário em processo de validação.
Traduzido e adaptado por Mónica Rocha – Estudante de Doutoramento. Colaboração das Terapeutas da Fala: Joana Caldas; Elsa Margarido; Margarida Morgado; Maria João Morgado. Orientação da Professora Doutora Joana Rato, PhD, UCP

Appendice 10

Sujeito:_____

Avaliação Global da Experiência da Gaguez pela pessoa – Versão Portuguesa -Adaptação para Professores-

Secção I: Informação Geral

Para cada item desta secção, faça um círculo à volta do número que se aplica ao/à seu/sua aluno/a. Pense nas experiências *atuais* e sentimentos do/a seu/sua aluno/a quando estiver a responder a cada questão. Se a questão não se aplicar ao/à seu/sua aluno/a, faça uma cruz na caixa e passe para a questão seguinte.

A. Informação geral sobre a fala do/a seu/sua aluno/a	**Sempre**	**Muitas Vezes**	**Às vezes**	**Raramente**	**Nunca**
1. Com que frequência o/a seu/sua aluno/a é capaz de falar fluentemente (sem gaguejar)?	1	2	3	4	5
2. Com que frequência a fala do/a seu/sua aluno/a lhe parece "natural"? (como a fala das outras crianças)	1	2	3	4	5
3. Com que frequência o/a seu/sua aluno/a usa as estratégias ou técnicas aprendidas na terapia da fala? (Se nunca teve terapia da fala, selecione "não teve/tem terapia da fala"	1	2	3 ☐ Não teve/tem terapia da fala	4	5
4. Com que frequência o/ seu/sua aluno/a diz *exatamente* o que quer dizer, mesmo que pense que possa gaguejar?	1	2	3	4	5

B. Na sua perspetiva, quanto o/a seu/sua aluno/a *sabe* sobre…?	**Imenso**		**Alguma coisa**		**Nada**
5. gaguez em geral	1	2	3	4	5
6. o que ajuda as pessoas a gaguejar *menos*	1	2	3	4	5
7 .o que faz as pessoas gaguejar *mais*	1	2	3	4	5
8. o que acontece à sua fala quando gaguejas	1	2	3	4	5
9. grupos de apoio ou de auto-ajuda para crianças se juntarem e falarem sobre gaguez (grupos de miúdos que se juntam e falam sobre gaguez)	1	2	3	4	5

C. De forma geral, na sua perspetiva, como o/a seu/sua aluno/a se sente em relação…?	**Muito Bem**	**Bem**	**Nem bem, nem mal**	**Mal**	**Muito Mal**
10. à sua fala, em geral	1	2	3	4	5
11. a falar com as pessoas, em geral	1	2	3	4	5
12. às técnicas ou ferramentas que aprendeu na terapia da fala (se nunca teve terapia da fala, selecione "não teve/tem terapia da fala")	1	2	3 ☐ Não teve/tem terapia da fala	4	5
13. a ser uma criança que gagueja	1	2	3	4	5
14. a ser chamado/a de gago/a pelas outras pessoas	1	2	3	4	5
15. a grupos de apoio ou de auto-ajuda para crianças que gaguejam	1	2	3	4	5

OASES-S – Overall Assessment of the speaker's Experience of Stuttering, Yaruss, S. & Quesal, R. (2010). Questionário em processo de validação.
Traduzido e adaptado por Mónica Rocha – Estudante de Doutoramento. Colaboração das Terapeutas da Fala: Joana Caldas; Elsa Margarido; Margarida Morgado; Maria João Morgado. Orientação da Professora Doutora Joana Rato, PhD, UCP

Appendice 11

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE
LISBOA·PORTO·VISEU

Declaração de consentimento informado

Eu, Mónica Filipa Soares Rocha, Doutoranda no programa de doutoramento em Ciências da Cognição e da Linguagem, Instituto de Ciências da Saúde da Universidade Católica Portuguesa, venho por este meio pedir autorização para avaliar o/a seu/sua educando/a através do preenchimento de escalas e realização de testes relacionados com a disfluência, temperamento, funções executivas e ansiedade. Solicito ainda a sua autorização para o preenchimento da escala: Avaliação Global da Experiência da Gaguez Pela Pessoa – Versão Portuguesa – adaptação para pais. Estes procedimentos estão incluídos no projeto de doutoramento intitulado: 'School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life' ('crianças portuguesas em idade escolar que gaguejam – desempenho socio-cognitivo e impacto da gaguez na sua qualidade de vida') , com a orientação da Professora Doutora Joana Rato (ICS-UCP) e professor Doutor Scott Yaruss (University of Pittsburgh).

Eu, _____, na qualidade de progenitor/a/ e/ou encarregado/a/ de educação de _____, declaro para os devidos efeitos, consentir a recolha e o tratamento de dados pessoais, bem como a utilização de gravação de imagem/áudio para fins unicamente científicos, assegurando-se a sua total confindencialidade e anonimato.

Declaro ainda, ter sido informado/a dos objetivos a que se propõe a utilização dos respetivos dados pessoais e de ter total liberdade para retirar o consentimento a qualquer momento.

_____

Encarregado/a de Educação

_____

Mónica Rocha

Estudante de Doutoramento

Appendice 12

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE
LISBOA·PORTO·VISEU

Declaração de consentimento informado

Eu, Mónica Filipa Soares Rocha, Doutoranda no programa de doutoramento em Ciências da Cognição e da Linguagem, Instituto de Ciências da Saúde da Universidade Católica Portuguesa, venho por este meio pedir autorização para avaliar o seu educando através do preenchimento de escalas e realização de testes relacionados com a disfluência, temperamento, funções executivas e ansiedade. Estes procedimentos estão incluídos no projeto de doutoramento intitulado: 'School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life' ('crianças portuguesas em idade escolar que gaguejam – desempenho socio-cognitivo e impacto da gaguez na sua qualidade de vida'), com a orientação da Professora Doutora Joana Rato (ICS-UCP) e professor Doutor Scott Yaruss (University of Pittsburgh).

Eu, _____, na qualidade de progenitor (a)/ encarregado (a) de educação de _____, declaro para os devidos efeitos, consentir a recolha e o tratamento de dados pessoais, bem como a utilização de gravação de imagem/áudio para fins unicamente científicos, assegurando-se a sua total confindencialidade e anonimato.

Declaro ainda, ter sido informado (a) dos objetivos a que se propõe a utilização dos respetivos dados pessoais e de ter total liberdade para retirar o consentimento a qualquer momento.

_____

Encarregado de Educação

_____

Mónica Rocha

Estudante de Doutoramento

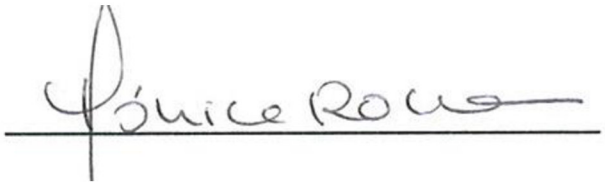
Appendice 13

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE
LISBOA·PORTO·VISEU

Carta de Apresentação

Eu, Mónica Filipa Soares Rocha, doutoranda no programa de doutoramento em Ciências da Cognição e da Linguagem, Instituto de Saúde da Universidade Católica Portuguesa, venho por este meio solicitar o preenchimento dos questionários que se encontram anexados a esta carta. Os questionários pretendem recolher informação sobre o/a seu/sua aluno/a que gagueja. Esta recolha de dados está a ser realizada no âmbito do projeto de doutoramento intitulado: 'School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life' ('Crianças portuguesas em idades escolar que gaguejam – Desempenho socio-cognitivo e impacto da gaguez na sua qualidade de vida'), com a orientação da Professora Doutora Joana Rato (ICS-UCP) e professor Doutor Scott Yaruss (University of Pittsburgh). Os questionários mencionados são: 1) Questionário de Caraterização Sociodemográfica (duas páginas); 2) Avaliação Global da gaguez pela pessoa – versão portuguesa – adaptação professores (quatro páginas). Salienta-se a importância de responder a **todas** as questões.

Os dados recolhidos serão analisados de forma anónima e a sua colaboração é voluntária. Será mantida a confidencialidade de toda a informação recolhida e a mesma será utilizada apenas nesta investigação.



Mónica Rocha

Estudante de Doutoramento

Appendice 14

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE
LISBOA·PORTO·VISEU

Pedido de Avaliação pela Comissão de Ética do ICS

Exmo presidente da Comissão da Ética do ICS,

Michel Renaud

Eu, Mónica Filipa Soares Rocha, Doutoranda no programa de doutoramento em Ciências da Cognição e da Linguagem, Instituto de Ciências da Saúde da Universidade Católica Portuguesa, venho por este meio pedir uma avaliação da Comissão de Ética do ICS do meu projeto de tese: 'School age portuguese children who stutter: Socio - cognitive performance and the impact of stuttering in their quality of life' ('crianças portuguesas em idade escolar que gaguejam – desempenho socio-cognitivo e impacto da gaguez na sua qualidade de vida'), com a orientação da Professora Doutora Joana Rato (ICS-UCP) e professor Doutor Scott Yaruss (University of Pittsburgh).
Junto envio o projeto de investigação bem como os instrumentos que serão utilizados no mesmo.

Mónica Rocha
Estudante de Doutoramento

Appendice 15

CATÓLICA
INSTITUTO DE CIÊNCIAS DA SAÚDE
LISBOA·PORTO·VISEU

INSTITUTO DAS CIÊNCIAS DA SAÚDE DA UCP
COMISSÃO DE ÉTICA

Parecer 34/2017

Projecto de Doutoramento em Ciências da Cognição e da Linguagem:

School Age Portuguese Children Who Stutter: Socio-cognitive Performance and the Impact of Stuttering in their Quality of Life

Doutoranda: Mónica Filipa Soares Rocha

Orientadores: Professora Doutora Joana Rato
Professor Doutor Scott Yaruss

Após boa recepção dos dois pedidos de aditamento relativos ao Projecto acima referido, pedidos incluídos no Parecer nº 29 de 24 de Abril, a Comissão de Ética do ICS da UCP considera que o Projecto de Investigação não levanta problemas de natureza ética.

Lisboa, 29 de Setembro de 2017

O Presidente da Comissão de Ética

Michel Renaud

Michel Renaud