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RESEARCH ARTICLE

Why Early Intervention is Essential in Preschool Age Children Who Stutter: A Systematic Review

Snijders S. ¹, Sevenants R. ¹, Eggers K. ^{1,2,3}, Van Eerdenbrugh S. ¹

¹ Dept. of Speech-Language Pathology, Thomas More University College, Belgium

² Dept. of Rehabilitation Sciences, Ghent University, Belgium

³ Dept. of Psychology and Speech-Language Pathology, Turku University, Finland

ABSTRACT

Background: For long, a wait-and-see approach was implemented for preschool age children who started to stutter. The last two decades, however, research indicated more and more that this is not the most appropriate action. Speech pathologists specialised in stuttering have gained much knowledge about stuttering near onset the past few years. A previous exploratory study showed that primary care professionals have not yet gained all this knowledge.

Aims: The aim of this systematic review was to fill the knowledge gaps of primary care professionals by formulating evidence-based information and to provide them to the primary care professionals.

Methods: To find evidence-based information for the knowledge that the respondents lack, a systematic literature search was conducted on 24 October 2022 in Cochrane, PubMed and Evidence Maps (American Speech-Language-Hearing Association). Systematic reviews and guidelines of the past decade about stuttering in preschool age children were consulted. Relevant information was selected. Resources were developed and fine-tuned by a focus group. Quality assessments were conducted on all included studies by using the Joanna Briggs Institute tools for systematic reviews and the Appraisal of Guidelines for Research and Evaluation (AGREE II) for guidelines. Findings are presented on a poster, flyer, website and video.

Results: Eight systematic reviews and one guideline met the inclusion criteria and were used to create information materials. Findings are that the cause of stuttering is very complex and influenced by multiple factors. Characteristics of stuttering include repetitions, prolongations, and blocks, but stuttering can also affect social and emotional functioning. It is clear that a wait-and-see approach is not the desired approach anymore.

Conclusions: The resources developed in this study can help primary care professionals in identifying stuttering and referring families of young children to a stuttering specialist, to maximize the chance on recovery for the child. A stuttering specialist will decide based on the stuttering characteristics and its development if a family needs advice and active follow-up or if treatment needs to be initiated.

Introduction

Stuttering is characterised by frequent or pervasive disruption of the normal rhythmic flow and rate of speech characterised by repetitions and prolongations in sounds, syllables, words, and phrases, as well as blocking and word avoidance or substitutions¹.

The prevalence of stuttering is 0.72%². The cumulative incidence under the age of six years is significantly higher than in later periods of life, with reported numbers of about 11.2% at the age of 4 years³. This means that stuttering recovers in a large proportion of children, either through spontaneous recovery or timely intervention. Recovery occurs in many children (50% - 94%)^{2,4}. These numbers differ due to different methodologies and different definitions for stuttering and recovery.

Stuttering can have a major impact on the life of the one who stutters and those around him or her⁵. Parents frequently report negative child behaviours caused by the stuttering, such as frustration, withdrawal, reduced or altered verbal output, avoidance behaviour... This indicates a level of awareness in young children who stutter. Even young children who do not stutter can develop negative thoughts towards children who stutter⁶. Stuttering can also affect social interactions between children in preschool⁷. Parents may also be concerned by their child's stuttering⁵. They can be protective, anxious, insecure or they may blame themselves.

It is therefore recommended that the social consequences of stuttering for the child and its environment are considered when making the decision not to initiate treatment immediately.

In the past, a 'wait and see' strategy was often applied. That is, not referring a child to a stuttering specialist (= a speech-language pathologist specialised in stuttering) because it was assumed that the stuttering would disappear. In the longitudinal study of Yairi et al.⁴, waiting for recovery of stuttering could take up to four years. In the first 18 months, only a minority of children recovered.

The wait-and-see approach is currently not the most appropriate approach, because (1) it is difficult to predict whether a child will recover spontaneously or not⁸, (2) early intervention seems to have a better outcome compared to no treatment⁹, (3) neuroplasticity is greater at this age and may explain treatment success at this age^{10,11}, and (4)

the social consequences of stuttering on children are even at preschool age not to be neglected^{12,13}.

When stuttering persists, negative consequences can also occur later in life. Adolescents who stutter are at higher risk of being bullied¹⁴. Stuttering in adulthood is associated with an increased risk of several anxiety disorders¹⁵.

In addition, not referring a child who needs therapy has more serious consequences than referring a child who, after further examination by a stuttering specialist, is found not to need therapy¹⁶. This is because a stuttering specialist will use evidence-based factors to consider the likelihood of recovery, such as gender, family history of stuttering and age at onset⁸.

For parents of young children who stutter, paediatricians are often the first point of contact¹⁷. Research has shown that general practitioners and paediatricians nowadays are less inclined to adhere to a wait-and-see strategy with young children who stutter than in the past¹⁶. However, unlike many other diagnoses, parental reporting of atypical behaviour about speech fluency did not result in a referral to a stuttering specialist. Thus, possibly not all young children who stutter are identified by primary care professionals. In the future, it is important that general practitioners and paediatricians are better informed about factors that may warrant referral to a stuttering specialist. Indeed, the willingness of physicians to refer young children who stutter depends on their knowledge about stuttering¹⁸. Many physicians believe that because most preschool children who stutter recover spontaneously (without speech therapy), referring young children to a stuttering specialist is not needed. The problem with this, however, is that children who do not recover have to wait longer for treatment, considerably reducing their chances on the most positive outcome.

Over the past 20 years, much new information about stuttering has appeared in the literature, including about the management of stuttering near onset. In response to this growing body of knowledge about stuttering near onset, Belgian primary care professionals were surveyed about their knowledge of stuttering in preschool age children and their referral behaviour. The respondents to the survey were general practitioners, paediatricians, and nurses and physicians of the organization Child & Family¹⁹. Child & Family follows the health evolution of Belgian infants from birth up to 30 months through regular, free consultations in regional offices. This includes a hearing screening after birth, a

vaccination program and advice about nutrition and general development. The primary care professionals were asked about their knowledge of stuttering, spontaneous recovery of stuttering, their referral behaviour, the information they received during their training and availability of informational resources. From this explorative study, it was clear that primary care professionals indicated a lack of knowledge, and they requested more information materials. The purpose of this research project was to develop information materials for these professionals with information about stuttering to guide them in identifying stuttering and implementing the appropriate referral actions. This paper describes on the systematic review procedure that was conducted to create the information materials.

Method

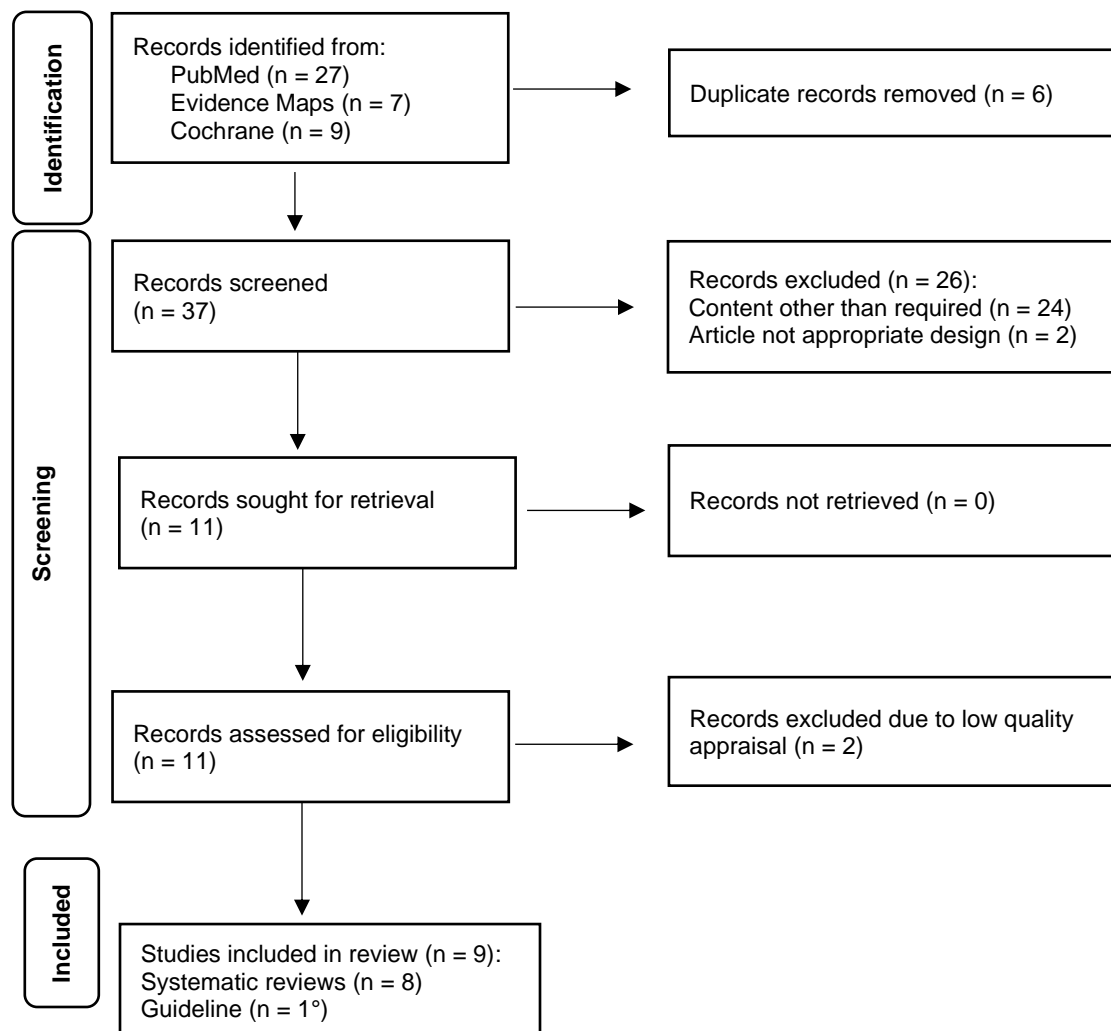
Previous exploratory study

An explorative study was conducted prior to this study. A questionnaire based on the questionnaire of Yairi and Ambrose²⁰ was completed by general practitioners (n=63), paediatricians (n=9) and nurses (n=82) and physicians (n=65) of Child & Family. Results were used for the systematic review of the research project.

Inclusion and exclusion criteria

Included in this review were guidelines and systematic reviews of the past decade (2013-2022). They had to contain information about the cause of stuttering, characteristics of stuttering and actions for stuttering near onset in preschool age children who stutter. Excluded were studies reporting other content and studies of poor methodological quality.

Figure 1: PRISMA Flowchart



^oThe guideline that was retrieved in Evidence Maps (ASHA) database, was not the most current version. Hence, the most current version was used instead^{21,22}.

Search strategy

The databases PubMed, Cochrane and Evidence Maps of the American Speech-Language-Hearing Association (ASHA) were consulted on 24 October 2022. In PubMed and Cochrane, the search string was based on (stutter* OR stammer* OR disfluent*) AND (child* OR preschool* OR pre-school* OR toddler OR developmental OR pediatri* OR kindergar*). In Evidence Maps, two predefined topics were chosen (“Fluency disorders”, “Early intervention”). All searches were limited to systematic reviews and guidelines published between 2013 and 2022.

The selection process was based on screening of title and abstract in a first stage, and on full text in a second stage. This was performed by the first author and confirmed by the last author. Inclusion criteria were also screened for all search results. Individual studies were selected if topics were not covered by the findings from the systematic reviews and guidelines. These were found through references in the guideline and through the knowledge of the authors.

The search procedure is visualised in the Prisma flow chart (Figure 1). The included systematic reviews and guidelines are presented in table 3.

Data extraction

Data were extracted from the included papers by the first author and checked by the fourth author. Data were collected about the cause of stuttering, the characteristics of stuttering and the actions for stuttering near onset.

Methodological quality assessment

The systematic reviews were appraised by the fourth author for their methodological quality, using the Joanna Briggs Institute tools²³. Table 1 shows the results. The guideline was appraised using the Appraisal of Guidelines for Research and Evaluation (AGREE II)²⁴ and was considered to be of acceptable quality. The selection process described in this paper was approved by the Centre for Evidence-Based Medicine (Cebam²⁵). This quality label is clearly presented on poster, website, flyer and video.

Table 1: Summary of risk of bias assessment of included systematic reviews

Checklist item	Baxter et al., 2015 ²⁶	Baxter et al., 2016 ²⁷	Bernard et al., 2022 ²⁸	Brignell et al., 2021 ²⁹	Johnson et al., 2016 ³⁰	Nye et al., 2013 ³¹	Sjøstrand et al., 2021 ³²	Sugathan et al., 2021 ³³
Is the review question clearly and explicitly stated?	+	+	+	+	+	+	+	+
Were the inclusion criteria appropriate for the review question?	+	+	+	+	+	+	+	+
Was the search strategy appropriate?	+	+	+	+	+	+	+	+
Were the sources and resources used to search for studies adequate?	+	+	+	+	+	+	+	+
Were the criteria for appraising studies appropriate?	+	+	+	+	+	+	+	+
Was critical appraisal conducted by two or more reviewers independently?	?	?	+	+	?	?	+	+
Were there methods to minimize errors in data extraction?	?	?	+	+	?	+	+	+
Were the methods used to combine studies appropriate?	+	+	+	+	+	+	+	+
Was the likelihood of publication bias assessed?	?	?	+	+	?	?	+	?
Were recommendations for policy and/or practice supported by the reported data?	+	+	+	+	+	+	+	+
Were the specific directives for new research appropriate?	+	+	+	+	+	+	+	+

Focus group

A focus group was organised to fine-tune the materials that were developed for the health care professionals. A first version of the poster and website were shown to a physician from Child & Family, a general practitioner, a stuttering specialist, a school care coordinator, a preschool teacher, a parent (of a child who has stuttered) and a parent of a preschool age child who does not stutter. The members of the focus group were all female. They gave verbal feedback and

formulated suggestions for the formulation of the information on the poster and website. This feedback and suggestions were incorporated in the final version of the informational materials.

Results

Previous exploratory study

Table 2 gives an overview of the responses on the questionnaire of the previous exploratory study.

Table 2: Overview results exploratory study

Topic	General practitioner (n = 62)	Paediatrician (n = 9)	Nurse Child & Family (n = 82)	Physician Child & Family (n = 65)
Contributes to onset of stuttering				
stress	93.5%	88.9%	86.6%	80.0%
emotional trauma	91.9%	100%	86.6%	80.0%
imitation of a peer who stutters	62.9%	88.9%	30.5%	23.1%
learnt behaviour	85.5%	77.8%	30.5%	20.0%
neurological deficit	79.0%	88.9%	68.3%	64.6%
Characteristics of stuttering				
blocking before a sound	96.7%	88.9%	87.8%	98.5%
repetition of a word	96.7%	88.9%	25.6%	41.5%
repetition of a sound	96.7%	88.9%	90.2%	92.3%
physical tension associated with speech	87.4%	77.8%	78.0%	66.2%
negative emotional reactions associated with speech	56.4%	77.8%	59.8%	75.4%
Action for stuttering near onset				
giving the advice to wait	72.5%	55.5%	36.6%	41.6%
not talking about it	16.1%	55.6%	43.9%	32.3%
referral to a stuttering specialist	64.5%	88.9%	35.4%	36.6%
Information				
received during education was insufficient	75.3%	87.5%	96.3%	96.9%
about stuttering in professional journals is insufficient	90.4%	87.5%	91.5%	83.1%

Three main topics were identified as being crucial for detecting stuttering and implementing appropriate referral behaviour, for which professionals gave various answers indicating a potential lack of knowledge.

In summary, most respondents (>80%) believe that emotions and stress contribute to the onset of stuttering. Less but still most respondents (64%) think that a neurological deficit underlies the onset of stuttering. Many respondents believe that imitation (between 23-89%) or learnt behaviour (between 20-85%) contribute to the onset of stuttering. It is clear that some misunderstanding about the onset of stuttering is present (e.g., stress and imitation are not causal factors) and clarification is needed.

Not all respondents (between 25-98%) recognize the visible features of stuttering; these are

repetitions, prolongations and blocking of sounds or syllables in speech. Less respondents (>56%), however, recognize negative emotional reactions as a characteristic of stuttering. This is a less visible feature but nonetheless an important feature of stuttering, also in preschool children.

Most general practitioners (72.5%) and paediatricians (55.5%) advise to take the wait-and-see approach, while less than half of the physicians and nurses of Child & Family (<42%) would do this. On the other hand, most general practitioners (64.5%) and paediatricians (88.9%) also refer children who stutter to a stuttering specialist, while less than half of the physicians and nurses of Child & Family would do this (<37%). Many respondents believe it is best not to talk about the stuttering. It is clear that not all health care

professionals take similar action. Clarification is needed.

Nearly all physicians (>75%) agreed that during their course of medicine or nursing, they were not sufficiently educated about stuttering in young children. They (>83%) also report that medical journals do not publish information about this disorder. The vast majority expressed an interest in a research publication on stuttering in young children in a professional journal. Among the physicians and nurses at Child & Family, more than 90% indicated that they have insufficient knowledge about stuttering. This study was performed to address this need.

Search results

The search yielded 43 recourses. After removing duplicates, 37 sources were screened. Of them, 28 sources were excluded due to other content than required, an inappropriate design or poor quality. Eventually, a total of nine studies met the inclusion criteria. Of these, eight were systematic reviews and one was a guideline. An overview of the literature process is presented in Figure 1. Table 3 gives an overview of the selected systematic reviews and guideline that were used to retrieve the information.

Table 3: The included systematic reviews and guideline containing information about stuttering near onset

Authors	Year	Title	Type of resource
Nederlandse Vereniging voor Logopedie en Foniatrie. Richtlijn stotteren bij kinderen, adolescenten en volwassenen ²²	2020	Richtlijn stotteren bij kinderen, adolescenten en volwassenen [Guideline stuttering in children, adolescents and adults].	Guideline
Baxter S, Johnson M, Blank L, Cantrell A, Brumfitt S, Enderby P, Goyder E ²⁶	2015	The state of the art in non-pharmacological interventions for developmental stuttering. Part 1: a systematic review of effectiveness	Systematic Review
Baxter S, Johnson M, Blank L, Cantrell A, Brumfitt S, Enderby P, Goyder E ²⁷	2016	Non-pharmacological treatments for stuttering in children and adults: a systematic review and evaluation of clinical effectiveness, and exploration of barriers to successful outcomes	Systematic Review
Bernard R, Hofslundsengen H, Frazier Norbury C ²⁸	2022	Anxiety and Depression Symptoms in Children and Adolescents Who Stutter: A Systematic Review and Meta-Analysis	Systematic Review
Brignell A, Krahe M, Downes M, Kefalianos E, Reilly S, Morgan A ²⁹	2021	Interventions for children and adolescents who stutter: A systematic review, meta-analysis, and evidence map	Systematic Review
Johnson M, Baxter S, Blank L, Cantrell A, Brumfitt S, Enderby P, Goyder E ³⁰	2016	The state of the art in non-pharmacological interventions for developmental stuttering. Part 2: qualitative evidence synthesis of views and experiences.	Systematic Review
Nye C, Vanryckeghem M, Schwartz JB, Herder C, Turner HM, Howard C ³¹	2013	Behavioural stuttering interventions for children and adolescents: a systematic review and meta-analysis.	Systematic Review
Sjøstrand Å, Kefalianos E, Hofslundsengen H, Guttormsen LS, Kirmess M, Lervåg A, Hulme C, Bottegaard Næss KA ³²	2021	Non-pharmacological interventions for stuttering in children six years and younger	Systematic Review
Sugathan N, Maruthy S ³³	2021	Predictive factors for persistence and recovery of stuttering in children: A systematic review.	Systematic Review

The cause of stuttering

The cause of stuttering is very complex. The guideline states that there are many theories about stuttering and that stuttering is considered a multifactorial disorder²². Multiple factors influence the development of stuttering. Insufficient information was found in the systematic reviews and guideline on this topic. Hence, individual studies were used to explain the cause of stuttering^{10,11,33,34-40}.

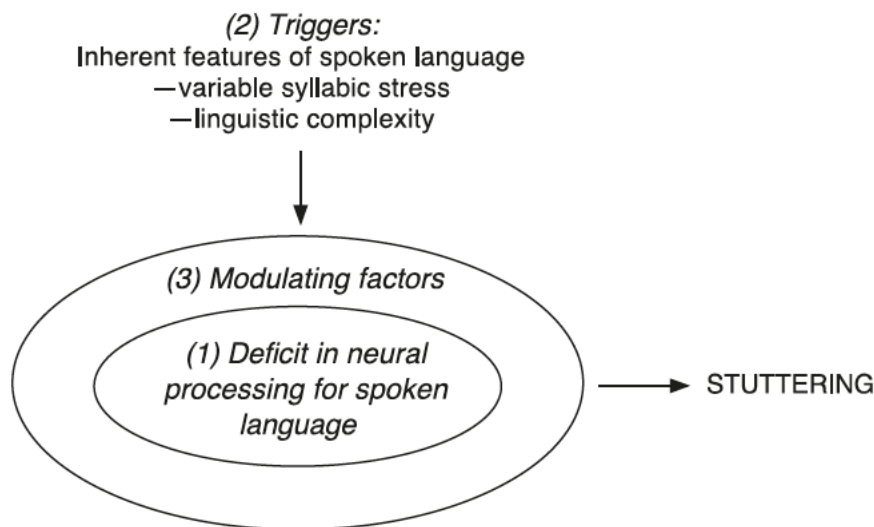
It has been calculated that 40-85% of the risk of developing stuttering can be explained by genetic background³⁴. Twin studies and family studies provide clear evidence for the existence of a strong genetic factor³⁴⁻³⁸. In some families, stuttering can be explained by a mutation in a single gene^{34,39}. In most cases, however, it is a complex condition caused by a combination of multiple hereditary factors.

Individual studies show anatomical differences in brain structures of children, especially of the grey and white matter^{10,11}. This indicates fewer nerve connections that provide information exchange in the brain. It is observed in young children who stutter. Even though more research is needed, nowadays neurological differences are generally considered to be the cause of stuttering²². Emotions

and stress are possible triggers of stuttering. There is no evidence that imitation or learnt behaviour contribute to the onset of stuttering³³.

One model to explain the onset of stuttering, is the Packman and Attanasio three factor model⁴⁰. The presence of a neural deficit for spoken language in all children who stutter is central to this model. That stuttering does not occur on each syllable can be explained by triggers that initiate stuttering at certain moments. According to Packman, triggers are associated with increased motor demands. An example is linguistic complexity. Speaking in shorter utterances and using simple vocabulary can decrease or control the stuttering. This triggering mechanism is "modulated by intrinsic factors" (p. 228), for example by physiological arousal (= the readiness of the body to react to stressful internal and external stimuli) or the available cognitive resources during communication. These modulating factors can change the threshold at which a moment of stuttering is triggered. That is, physiological arousal may increase with excitement and hence the stuttering increases. Also, cognitively more demanding tasks may trigger stuttering more quickly than in speaking situations without cognitive demands.

Figure 2: The Packman and Attanasio 3-factor model of moments of stuttering



Source: Packman (2012). Theory and therapy in stuttering: A complex relationship. *Journal of Fluency Disorders* 37 (2012) 225–233. Figure 1, p. 227

Characteristics of stuttering

Normal disfluencies occur in all preschool age children and are part of normal speech-language development⁴. A typically developing child will sometimes repeat words, but these do not really affect the child's fluency of speech. It is normal for

children who are thinking while producing a sentence to interject words such as 'uhm' in their speech.

Preschool age children who stutter produce normal disfluencies but also have stuttering moments. These include repetitions of sounds and syllables,

prolonged and blocked sounds²². A reliable cut-off to diagnose children who stutter and those who don't is three syllables characterized by repetitions of a sound, syllable or word, sound prolongations or blocks per 100 produced syllables^{4,32}. Because stuttering is variable, stuttering can be less or more severe in the same child in different situations. Therefore, caution with the cut-off is needed: a child who only has one stuttered disfluency on 100 syllables can also be a child who stutters.

Sometimes young children exhibit secondary behaviours associated with their stuttering, such as blinking their eyes or bringing their hands to their mouth, indicating awareness and possible fighting against the stuttering moments.

Four systematic reviews reported that stuttering can also affect social and emotional functioning^{26,28,32,30}. Some children may experience or develop feelings such as anxiety or embarrassment because of their stuttering.

Action for stuttering near onset

In the previous century, the wait-and-see approach was usually advised because it was reported that spontaneous recovery occurs in many children⁴. Nowadays however, research clearly shows that recovery or persistence of stuttering cannot be predicted⁸. Stuttering specialists evaluate several factors over time and make a facts-based decision about what is known to be predictive for recovery or persistence of stuttering and decide whether treatment is necessary or not. If not, active monitoring is suggested. That is, giving advice to the family and following up the child and family at regular times. In the study reporting recovery⁴, families received advice and applied that advice in their daily conversations with their child before recovery was reported. Therefore, it has become clear that professional intervention is essential as a catalytic agent to obtain recovery.

Systematic reviews about treatment effect in preschool children indicate that treatment is successful in most preschool children who stutter^{22,26,27,29,31,32}. One individual study in

particular⁹, showed a statistically significant effect on the frequency of stuttering moments in the speech in a group of children who stuttered and received treatment compared to a group of children who stuttered and did not receive treatment for 9 months. This finding is of utmost importance to show that intervention at this age affects the development of stuttering. Knowing that children, even at preschool ages, often suffer from lower quality of life or negative emotions because of their stuttering^{12,13}, a wait-and-see approach is clearly not the desired approach anymore.

In addition to timely referral to a speech language therapist, it is a good idea to talk about the stuttering with the child^{27,30}. It is important, because young children are even at this age often aware of stuttering and may experience negative emotions^{12,13}. It helps to listen actively to the child and to be non-judgemental, patient, and caring during a conversation²⁷. This will build feelings of confidence, acceptance, understanding and trust.

Informational materials and focus group

The information above was used to create a poster, website, flyer and video. They contain information about the cause of stuttering, the characteristics of stuttering and the actions near onset of stuttering. A first version of the poster and website were presented to a focus group. The main outcomes of this focus group were that three versions of the website needed to be developed: one for medical professionals (e.g., general practitioners, paediatricians, nurses), one for schools (e.g., preschool teachers, care coordinators), and one for parents of preschool children. These versions should contain the same information but sequenced differently. For example, for physicians it is important to have the information about referral straight away. The focus group also provided advice on the language used on the materials. They suggested to use pictograms to increase the accessibility of the information. Figure 3 displays the final version of the flyer.

Figure 3: Flyer

Stuttering in preschoolers



Cause

The cause of stuttering is very complex. In most children it is explained by the processing of speech that is different in the brain and by genetic factors.



Characteristics

Visible features are repetitions of sounds or words and being stuck in or before a sound. Stuttering also has an impact on social and emotional functioning. Sometimes children show secondary behaviours such as gasping for breath, which indicates fighting the stuttering.



When do we speak of stuttering?

When there is an increased presence of abnormal disfluencies, it is appropriate to speak of stuttering.



How can we help?

- Take the time to talk to the child.
- Be relaxed and look at the child while you listen.
- Wait your turn to speak and do not interrupt the child.
- Respond to what the child says, not to how it was said.



Talking about the stuttering

It is important that the child and his/her environment acknowledge their thoughts and feelings and make stuttering a subject for discussion. The child is often already aware of the stuttering. By talking about it, the child will worry less. Ask a speech therapist specialized in stuttering for specific tips.



Referral

Do you think a child stutters? Do not wait. Parents should contact a speech therapist specialized in stuttering. With the right advice or treatment, the chance to recover before the age of 6 years is greater.



More information

<https://www.thomasmore.be/stotteren-bij-kleuters>



Researchers & authors: Diede Piedfort, Steffi Snijders, Renke Sevenants, Kurt Eggers & Sabine Van Eerdenbrugh



Discussion

Not all primary care professionals are up to date with the recent knowledge about stuttering in preschool age children. This affects their referral behaviour. In addition, these professionals indicate that they did not receive sufficient knowledge about stuttering during their training and show interest in informational materials.

Recent research shows that physicians' willingness to refer young children who stutter to a stuttering specialist depends on their knowledge about stuttering¹⁶. This is consistent with the findings of the previous exploratory study. Answers from the general practitioners, paediatricians, and nurses and physicians of Child & Family show that not all of them are up to date with the recent evidence. The professionals especially lack knowledge about the

cause of stuttering, the characteristics of stuttering and the actions near onset, such as the timely referral to a speech language therapist specialised in stuttering.

First, many respondents believe that emotions, stress, and imitation or learnt behaviour contribute to the onset of stuttering. The literature review shows that multiple factors influence the development of stuttering²². Nowadays a neurological deficit is generally considered to be the cause of stuttering. Emotions and stress are possible triggers of stuttering. There is no evidence that imitation or learnt behaviour contribute to the onset of stuttering³³.

Additionally, nearly all respondents recognize the visible features of stuttering. Less respondents recognize negative emotional reactions as a characteristic of stuttering, whereas the literature shows that stuttering can also affect social and emotional functioning^{26,28,30,32}.

Finally, quite a large group does not give any action to the stuttering at first but apply the wait-and-see approach. This is not consistent with the advice from the literature review: A timely referral of preschool age children who stutter is crucial for several reasons^{22,26,27,29,31,32}.

The preliminary, explorative survey revealed the need for information materials about stuttering in preschool age children. A poster and flyer in English are the result of this research project, the website and an information video are translated and freely available. The evidence-based method that supported the development of these information materials and the easy and free access of the materials are of great importance to the authors of this study. Hopefully this results in more timely referrals, adequate and timely intervention, and greater numbers of recovery before the age at which stuttering is likely to become persistent (start of primary school, 6 years).

Involving stakeholders in the development of the materials is a step in the evidence-based approach which translated the research findings to the daily, clinical practice. The focus group indicated that the poster and website are useful for providing information. The participants gave suggestions to improve the practicality of the materials. They recommended working with pictograms to increase access to non-native speaking families. The pictograms visually support the text.

Strengths and limitations

The strength of this study is that information materials were developed tailored to the needs of primary care professionals and based on recent scientific insights. An exploratory study made it possible to identify specifically which knowledge was lacking.

It is an advantage that there are multiple forms of the information material, for example, a concise overview can be obtained by viewing the poster, flyer, or video. The poster can be printed in the size that is required. Those who want more extensive information can consult the website. Everyone who regularly meets preschool age children can get valuable information from these materials.

The information for primary care professionals was certified by Cebam, which counts as a quality label in Belgium²⁵. It guarantees the methodological quality of the resources.

Limitations of the study include that certain topics could not be underpinned by scientific information of a systematic review or guideline, but only by findings from individual studies. Also, the group of paediatricians in the exploratory study was small compared to the other groups. Another limitation is the inclusion of studies with a higher risk of bias^{26,27} in some systematic reviews. Certain reviews have a limited number of studies^{28,31}. Not every review captured all available studies, because of specific words in the search string or because of excluding certain study designs.

Implications for practice and future research

The developed information resources can be used by primary care professionals such as general practitioners, paediatricians and professionals who follow the evolution of young children the first few years after they are born. These professionals can find up-to-date, evidence-based information about detecting stuttering near onset and referring behaviour. It is of utmost importance that families of young children who started to stutter consult a stuttering specialist timely (that is, soon after onset), to receive the most appropriate intervention, whether that is limited to individually tailored advice or initiating treatment.

In the meantime, an awareness campaign was set up to provide the materials to Belgian primary care professionals in training and in the practice. Future research could investigate whether knowledge among professionals has increased and actions have changed.

Conclusion

The wait-and-see approach was considered an appropriate approach for preschool age children for a long time. With the research findings of the last two decades, this approach does not seem to be the best approach. Waiting for recovery of stuttering may take up to four years after onset, and in the first 18 months only a minority of children recover. Reasons for the switch of approach include the risk on developing negative feelings about stuttering, the fact that recovery cannot be predicted and the fact that treatment at preschool age is most successful compared to stuttering in older children.

Primary care professionals indicate that they are not up to date with the recent knowledge about stuttering in preschool age children. Subsequent

behaviour may therefore not be the most appropriate for a child who stutters near onset. The development of a poster, flyer, website and information video can contribute to a better knowledge about stuttering and timely referral of primary care professionals to a stuttering specialist.

Conflict of interest statement

The authors have no conflicts of interest to declare.

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References

1. World Health Organization. International statistical classification of diseases and related health problems (11th ed.). 2019. <https://icd.who.int/>
2. Yairi E, Ambrose N. Epidemiology of stuttering: 21st century advances. *Journal of Fluency Disorders*. 2013;38(2):66-87.
3. Reilly S, Onslow M, Packman A, Cini E, Conway L, et al. Natural history of stuttering to 4 years of age: A prospective community-based study. *Pediatrics*. 2013;132(3):460-467.
4. Yairi E, Ambrose NG. Early childhood stuttering I: Persistency and recovery rates. *Journal of Speech, Language, and Hearing Research*. 1999;42(5):1097-1112.
5. Langevin M, Packman A, Onslow M. Parent perceptions of the impact of stuttering on their preschool age children and themselves. *Journal of Communication Disorders*. 2010;43(5): 407-423.
6. Weidner ME, St. Louis KO, Burgess ME, LeMasters SN. Attitudes toward stuttering of nonstuttering preschool and kindergarten children: A comparison using a standard instrument prototype. *Journal of Fluency Disorders*. 2015;44:74-87.
7. Langevin M, Packman A, Onslow M. Peer responses to stuttering in the preschool setting. *American journal of speech-language pathology*. 2009;18(3):264-76.
8. Singer CM, Hessling A, Kelly EM, Singer L, Jones RM. Clinical characteristics associated with stuttering persistence: A meta-analysis. *Journal of Speech, Language, and Hearing Research*. 2020;15;63(9):2995-3018.
9. Jones M, Onslow M, Packman A, Williams S, Ormond T, Schwarz I, GebSKI V. Randomised controlled trial of the Lidcombe programme of early stuttering intervention. *BMJ*. 2005;22;331(7518):659-664.
10. Chang SE, Garnett EO, Etschell A, Chow HM. Functional and neuroanatomical bases of developmental stuttering: current insights. *The Neuroscientist*. 2019;25(6):566-582.
11. Chang SE, Zhu DC, Choo AL, Angststadt M. White matter neuroanatomical differences in young children who stutter. *Brain*. 2015;138(3):694-711.
12. McAllister, J. Behavioural, emotional and social development of children who stutter. *Journal of Fluency Disorders*. 2016;50:23-32.
13. Briley PM, O'Brien K, Ellis C. Behavioural, emotional, and social well-being in children who stutter: Evidence from the National Health Interview Survey. *Journal of Developmental and Physical Disabilities*. 2019;31:39-53.
14. Blood GW, Blood IM. Bullying in adolescents who stutter: Communicative competence and self-esteem. *Contemporary Issues in Communication Science and Disorders*. 2004;31(Spring):69-79.
15. Iverach L, O'Brian S, Jones M, Block S, Lincoln M, Harrison E, et al. Prevalence of anxiety disorders among adults seeking speech therapy for stuttering. *Journal of Anxiety Disorders*. 2009;23(7):928-34.
16. Winters KL, Byrd CT. Pediatrician referral practices for children who stutter. *American Journal of Speech-Language Pathology*. 2020; 29(3): 1404-1422.
17. Perez H, Stoeckle J. Stuttering: Clinical and research update. *Can Fam Physician*. 2016;62(6): 479-484.
18. Lees R, Stark C, Baird J, Birse S. Primary care professionals' knowledge and attitudes on speech disfluency in pre-school children. *Child Language Teaching and Therapy*. 2000;16(3): 241-254.
19. Child & Family (Kind & Gezin). 2022. <https://www.kindengezin.be/nl>
20. Yairi E, Carrico DM. Early childhood stuttering: Paediatricians' attitudes and practices. *American Journal of Speech-Language Pathology*. 1992;1(3):54-62.
21. Pertijs, MAJ, Oonk, LC, Beer, de JJA, Bunschoten, EM, Bast, EJEG, Ormond, van J, Rosenbrand, CJGM, Bezemer, M, Wijngaarden, van LJ, Kalter, EJ, Veenendaal, van H. Evidence-based Richtlijn Stotteren bij kinderen, adolescenten en volwassenen. *Nederlandse Vereniging voor Logopedie en Foniatrie*, Woerden. 2014.
22. Nederlandse Vereniging voor Logopedie en Foniatrie. Richtlijn stotteren bij kinderen, adolescenten en volwassenen. 2020. <https://www.nvlf.nl/kennis/inhoudelijke-richtlijnen/>
23. Joanna Briggs Institute tools. 2022. <https://jbi.global/critical-appraisal-tools>
24. AGREE Next Steps Consortium. Appraisal of Guidelines for Research & Evaluation (AGREE) II Instrument. <https://www.agreetrust.org/>
25. Cebam. 2022. <https://www.cebam.be/>
26. Baxter S, Johnson M, Blank L, Cantrell A, Brumfitt S, Enderby P, Goyder E. The state of the art in non-pharmacological interventions for developmental stuttering. Part 1: a systematic review of effectiveness. *International journal of language & communication disorders*. 2015;50(5):676-718.
27. Baxter S, Johnson M, Blank L, Cantrell A, Brumfitt S, Enderby P, Goyder E. Non-pharmacological treatments for stuttering in children and adults: a systematic review and evaluation of clinical effectiveness, and exploration of barriers to successful outcomes. *Health Technol Assess*. 2016;20(2):1-302,v-vi.

28. Bernard R, Hofslundsengen H, Frazier Norbury C. Anxiety and depression symptoms in children and adolescents who stutter: A systematic review and meta-analysis. *Journal of Speech, Language, and Hearing Research*. 2022;65(2):624-44.
29. Brignell A, Krahe M, Downes M, Kefalianos E, Reilly S, Morgan AT. A systematic review of interventions for adults who stutter. *Journal of Fluency Disorders*. 2020;64:105766.
30. Johnson M, Baxter S, Blank L, Cantrell A, Brumfitt S, Enderby P, Goyder E. The state of the art in non-pharmacological interventions for developmental stuttering. Part 2: qualitative evidence synthesis of views and experiences. *International Journal of Language & Communication Disorders*. 2016;51(1):3-17.
31. Nye C, Vanryckeghem M, Schwartz JB, Herder C, Turner III HM, Howard C. Behavioural stuttering interventions for children and adolescents: A systematic review and meta-analysis. *Journal of Speech, Language and Hearing Research*. 2013;56(3):921-932.
32. Sjøstrand Å, Kefalianos E, Hofslundsengen H, Guttormsen LS, Kirmess M, Lervåg A, Hulme C, Bottegaard Næss KA. Non-pharmacological interventions for stuttering in children six years and younger. *Cochrane Database Syst Rev*. 2021;9(9):CD013489.
33. Sugathan N, Maruthy, S. Predictive factors for persistence and recovery of stuttering in children: A systematic review. *International Journal of Speech-Language Pathology*. 2021;23(4):359-371.
34. Van Beijsterveldt CE, Felsenfeld S, Boomsma DI. Bivariate genetic analyses of stuttering and nonfluency in a large sample of 5-year-old twins. *Journal of Speech, Language, and Hearing Research*. 2010;53(3):609-19.
35. Bloodstein O, Ratner NB. A handbook on stuttering. Clifton Park, NY: Thomson/Delmar Learning; 2008.
36. Dworzynski K, Remington A, Rijdsdijk Frühling, Howell P, Plomin R. Genetic etiology in cases of recovered and persistent stuttering in an unselected, longitudinal sample of Young Twins. *American Journal of Speech-Language Pathology*. 2007;16(2):169-78.
37. Fagnani C, Fibiger S, Skytthe A, Hjelmborg JV. Heritability and environmental effects for self-reported periods with stuttering: A twin study from Denmark. *Logopedics Phoniatrics Vocology*. 2010;36(3):114-20.
38. Frigerio Domingues CE, Grainger K, Cheng H, Moretti-Ferreira D, Riazuddin S, Drayna D. Are variants in sex hormone metabolizing genes associated with stuttering? *Brain and Language*. 2019;191:28-30.
39. Kang C, Riazuddin S, Mundorff J, Krasnewich D, Friedman P, Mullikin JC, et al. Mutations in the lysosomal enzyme-targeting pathway and persistent stuttering. *New England Journal of Medicine*. 2010;362(8):677-85.
40. Packman A. Theory and therapy in stuttering: A complex relationship. *Journal of Fluency Disorders*. 2012;37(4):225-33.