Clinical Guideline

'Stuttering in Children, Adolescents and Adults'

Colofon: *Clinical Guideline Stuttering in Children, Adolescents and Adults*

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The CBO, was based in Utrecht (1979-2015), aimed to support the professional associations and health care organizations of individual practitioners in improving patient care. The CBO offered this through programs and projects which supported and assisted with the systematic and structured, improvement and maintenance of quality patient care.

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Summary of the Recommendations

1. What are the Indicators to Treat Children, Adolescents and Adults who Stutter?

Stuttering is a disorder in the fluency of speech. The first symptoms are usually observed in the first years of life, shortly after language development has started. Stuttering can develop into a communication problem that may severely impair social development and personal well-being. Stuttering may be persistent and difficult to treat at later ages. There is evidence from clinical studies that early interventions enhance the chances of recovery. However some children who start to stutter may recover without any intervention. There is currently insufficient clarity about the number of children who recover spontaneously and it is still not possible to simply predict which children will recover naturally. Recommendations will be formulated to enable speech & language therapists and fluency specialists to take evidence-based decisions about at which point to start treating the person who stutters or to allow spontaneous recovery to take place (see Chapter 3).

Quality of evidence

Spontaneous recovery and the *effectiveness of treatment* play an important role in providing advice regarding the question of when a person who stutters needs to be treated. Figures about spontaneous recovery show large differences and risk factors for persistent stuttering have only been investigated to a limited extent.

Treating children before age six has a significant effect on the frequency of stuttering and there is a reasonable certainty that the estimate of this effect size corresponds to the actual effect size. Treatment of older children, adolescents and adults is less effective in reducing the percentage of stuttered syllables. There are insufficient data available to determine how effective treatment of stuttering is on outcome measures such as quality of life, avoidance behaviour, participation and naturalness of speech.

Monitoring

Monitoring is an active process where the speech & language therapist or fluency specialist systematically follows the progress of the stuttering. After the diagnosis / consultation, the speech & language therapist or fluency specialist will train parents in the focused observation and recording of the child's stuttering symptoms. The speech & language therapist or fluency specialist will continue training the parents until they reach an agreement about the severity of the stuttering observed. The use of recording system to be used, the frequency of recording, as well as the mode and frequency of reporting the recorded data back to the speech & language therapist or fluency specialist will be discussed and agreed in consultation with the parents. During the monitoring process the speech & language therapist or fluency specialist will check whether the mode of assessing stuttering severity by the parents continues to agree with his/her own assessment. A review

assessment will be undertaken at least once every six months. These review assessments should preferably take place six months and one year after the child has started to stutter.

Recommendations

The recommendations below should be considered in their entirety.

1. The treatment of children who begin to stutter before the age of four, starts before they become five years of age.
2. The speech & language therapist or fluency specialist will monitor the child who started to stutter, before four years of age, for signs of spontaneous recovery during a period of one year after onset.
3. The speech & language therapist or fluency specialist will start treatment 12 months after onset, when the severity of stuttering has not clearly* decreased in the last 3-6 months.
4. The speech & language therapist or fluency specialist will take into account the recovery pattern of stuttering in the family, gender, phonological skills, additional disorders and the temperament of the child in the decision to start intervention or to monitor fluency development.
5. The speech & language therapist or fluency specialist will start intervention (even if the child has been stuttering for less than 12 months) if the child suffers from stuttering, if the parents are concerned about the child's stuttering or if the child is showing reluctance to communicate.
6. Parents of a child under 6 years of age fill in the Stuttering Screening List (SSL) to determine whether further diagnostic assessment by the speech & language therapist or fluency specialist is indicated.
7. The speech & language therapist or fluency specialist will inform the medical professionals, teachers and supervisors at day care centers within his/her network about the purpose and usage of the SSL and he/she will provide information about relevant websites.
8. In cases of children over 6 years of age, adolescents or adults, seeking help in stuttering, the speech & language therapist or fluency specialist will, together with the client, make an inventory of ICF-elements (International Classification of Functioning, Disabilities and Health) to find out whether treatment is indicated.

* a clear decrease in stuttering corresponds to an average reduction of at least 2 points on the 8-point stutter severity scale (Yairi & Ambrose, 2005) or an average reduction of 2 points on the Lidcombe Program stuttering severity scale. This decrease in severity needs to be observed by both the parents and the speech & therapy therapist / fluency specialist.

Rationale for the recommendations

- Stuttering may have severe consequences on the quality of life of children and adults who stutter.
- Treating young children who stutter is more effective than treating children who are six or older: the risk of a recurrence is smaller and the chance of persistent recovery is greater (Ingham & Cordes, 1998; Lincoln et al., 1996; Koushik et al., 2009).

- Treating stuttering within the 15 months after onset, has a higher chance of recovery than starting treatment once the stuttering has been present for more than 15 months (Ingham & Cordes 1998).
- Observing the first symptoms of stuttering after the age of 4 or 5 has been associated with an increased risk of persistent stuttering (Yairi & Ambrose 2005; Seery & Yairi 2011).
- A large group of children who start to stutter will recover fully without treatment. However, there are large differences in the figures for spontaneous recovery (Yairi & Ambrose 2013; Onslow & O'Brian 2013).
- Delaying treatment for a year does not extend the length of treatment required (Jones et al., 2000; Klingston et al., 2003).
- The risk of persistent stuttering increases if stuttering is still present one year after the first symptoms have been observed (Yairi & Ambrose 2005; Seery & Yairi 2011).
- A downward trend in stutter severity during the first year after the first stuttering symptoms have been observed is a clear sign of a potential recovery.
- A reduction in the number of repetitions, the number of units per repetition, blocks and extensions and additional physical behaviours, as well as a reduction in the rate of repetitions are positive predictors of recovery (Yairi & Ambrose 2005; Seery & Yairi 2011).
- The severity of stuttering in the first year is not a predictor of persistent stuttering or predictive of a recovery in stuttering (Yairi & Ambrose, 2005; Seery & Yairi, 2011).
- Stutter severity, the presence of secondary behaviour, blocks and extensions are predictors of persistent stuttering if the stutter has been present for a year or more (Yairi & Ambrose 2005; Seery & Yairi 2011).
- Therapeutic intervention is indicated if the parents / carers are concerned (Reilly et al., 2013).
- The average weighted SLD score decreases by more than one half in the group of children who recover from stuttering in the six months to a year after stuttering has started. The average weighted SLD score decreases minimally in the group of children who continue to stutter in the six months to a year after stuttering has started. A comparable trend is evident for stutter severity (Yairi & Ambrose, 2005).
- If persistent stuttering is present in the family of the child who stutters then the chance of the child continuing to stutter is 65%. The presence of recovered stuttering in the child's family predicts that the chance of persistency decreased to about 35% (Yairi & Ambrose, 2005; Seery & Yairi, 2011). The ratio of boys to girls who stutter at a young age is 2:1. The ratio of men to women who stutter in adults is approximately 4:1. Girls recover more quickly. If there is no clear recovery observable in a girl who has been stuttering for more than one year then the chance of persistent stuttering increases (Yairi & Ambrose 2005; Seery & Yairi 2011).
- Additional disorders, a higher negative reaction and lower self-regulation may complicate the stuttering problems and increase the chance of persistent stuttering.
- Parents of children who stutter regularly report that they were referred too late to a speech & language therapist or fluency specialist.
- The SSL is the most commonly used screening instrument in the Netherlands. The SSL is freely available via various websites.

2. What is the Diagnostic Value of Tests for Children and Adults who Stutter?

Stuttering is considered to be a multifactorial problem. Diagnosing stuttering is more than just establishing whether or not stuttering is present. During the assessment phase, the speech & language therapist needs to explore the various ICF domains (functions, external factors, contextual factors, personal factors, participation and activities) in more depth and detail, then analyse and interpret their mutual associations. In order to realise as complete a picture as possible, the speech & language therapist will use the best available diagnostic instruments that are applicable and will draw in all the individuals from the client's environment who are relevant to the diagnostic process. The value of the instruments most commonly used in the Netherlands is discussed in this chapter.

Conclusion about the diagnostic value of tests

The validity and reliability of a number of instruments that are in use or of interest in the Netherlands will be discussed (see chapter 4). The following instruments will be covered:

- **Stuttering Screening List** and **Stuttering Detection Instrument**, both used to flag / detect stuttering
- **Communication Attitude Test -DR**,
- **Test for Stuttering Severity (TfS) readers / non-readers** and **Stuttering Severity Instrument** (SSI- 3/4), both used for determining the severity of *stuttering*
- **Behaviour Assessment Battery (BAB)** including **Communication Attitude Test** and **Erickson-S24** and **Overall Assessment of Speaker's Experience of Stuttering (OASES)**, both used to detail various aspects of the *quality of life*.

TfS, SSI-4, OASES and BAB also measure various aspects of *avoidance behaviour*.

Quality of evidence

No single instrument has had every aspect of validity and reliability investigated. This means that there is no fully proven, valid and reliable test available with adequate standardisation. At this point the BAB (children and adults) and the SSI appear to be the most valid and reliable instruments.

Values and preferences

a. People who stutter

Benefits of the SSI 3/4:
- places little burden on Persons Who Stutter (PWS) with a minimal time investment;
- it can be used both within and beyond practice settings.

Disadvantages of the SSI 3/4:
- The SSI 3/4 is an English-language test, there is no Dutch translation available.

Disadvantages of the BAB (adults and children):
- it is more time intensive;
- it contains Flemish expressions;
- it requires a good command of Dutch.

b. Speech & language therapist / fluency specialists

Benefits of the SSI 3/4:
- it is used internationally;
- it can be used both within and beyond practice settings.

Disadvantages of the SSI 3/4:
- The SSI 3/4 is an English-language test, there is no Dutch translation available.

Benefits of the BAB:
- it has been investigated the most thoroughly and scores well on various aspects of validity and reliability.
- it fits in well with the ICF framework.

Disadvantages of the BAB (adults and children):
- it is more time intensive;
- it contains Flemish expressions;
- it requires a good command of Dutch.

The OASES provides options for the future, but was not available in the Netherlands in 2014.

c. Costs

A disadvantage of the Behaviour Assessment Battery (BAB) for children and adults is the high purchase price.

Recommendations

The starting point for the recommendations below is that the speech & language therapist works in accordance with the professional standards for clinical decision making and outcome

measurement. It is assumed that the symptoms and presentation have been explored and that a case history interview has been undertaken with the PWS and/or parents/carers.

9. The speech & language therapist or fluency specialist determines stuttering severity with a suitable instrument. The Stuttering Severity Instrument (SSI) is preferred because the SSI is used internationally, is valid and seems to be reliable.
10. The speech & language therapist or fluency specialist assesses functions, external factors, contextual factors, individual factors, participation and activities according to the ICF principles during diagnostic assessment. The Behaviour Assessment Battery is recommended as a diagnostic instrument for children over 6 years, adolescents and adults. For children under 6 years of age no specific recommendation is given due to the lack of specific diagnostic instruments for this age group.

Rationale for the recommendations

- A relatively large weight has been given to the validity, reliability, availability and the link with international developments, and less to factors such as costs, time investment, and the language of the BAB and the English version of the SSI.

3. What are the Effects of Stuttering Therapy in Children who Stutter up to Age Six?

The effectiveness of the Demands and Capacities Model (DCM) and the Lidcombe Program (LP) will be described in Chapter 5. In the Netherlands the DCM and the LP are the most commonly employed interventions for treatment of stuttering in children up to age 6. The effectiveness of Speech Motor Training (Riley & Riley, 1999) and Social-Cognitive behavioural therapy (Boey, 2003) will also be discussed to supplement the conclusion and recommendation.

The starting point is that the effectiveness of a stuttering therapy has to be evaluated in a randomised controlled trial (RCT) in intervention studies with a control group. Both studies where a stuttering therapy is compared to "doing nothing", as well as studies comparing various stuttering therapies will be considered. Desired and undesired effects of stuttering therapies are evaluated in terms of:
- stuttering severity;
- avoiding speaking situations (secondary behaviours);
- participation;
- quality of life;
- naturalness of speech.

For the chapters about the effectiveness of stuttering therapy and pharmacotherapy (Chapters 6-7-8), the same principles and outcome measures are used.

Recommendation

> 11. Discuss with the parents of children up to 6 years of age who stutter the benefits of stuttering treatment versus no treatment option (reduction of percentage stuttered syllables and stuttering severity). Explain the differences between the 'Lidcombe program' and treatment based on the 'Demands and Capacities Model'. Come to a shared decision with the parents which of the two approaches is preferred.

Rationale for the recommendation

- There is a reasonable degree of certainty that treating children who stutter up to age 6 with the Lidcombe Program is more effective in reducing the percentage of stuttered syllables compared to no treatment.
- Harmful effects of treatment have not been found for the Lidcombe Program.
- There is an increasing degree of trust that the effect of treatment with the Lidcombe Program is no more effective than treatment based on the Demands and Capacities Model and vice versa.
- Differences in effects and costs between the LP and DCM treatment are small. The LP is a good alternative to DCM treatment in Dutch primary care.
- The Lidcombe Program and the Demands and Capacities Model should take preference above treatment with Speech Motor Training and Social Cognitive Behavioural Therapy given the degree of certainty regarding the size of the treatment effect.

4. Effectiveness of Stuttering Therapies in Children Aged Between 6 and 13

The effectiveness of stuttering therapies for children aged between 6 and 13 is discussed in Chapter 6.

Recommendation

> 12. The treatment of stuttering in children between the ages of six and thirteen years should be based on a treatment plan that contains all ICF elements and focuses on the types of behaviors, emotions and cognitions that have been identified, in collaboration with the child who stutters and his parents during assessment.

Rationale for the recommendation

- There is a (very) limited degree of certainty regarding the extent to which the number of stuttered syllables is reduced in children aged 6 to 13 after treatment.
- There is insufficient evidence that one stuttering therapy is more effective than another in the treatment of children who stutter aged 6 to 13.

- There is some but limited degree of certainty that a reduction in stuttering severity in children aged 6 to 13 is less than in children up to age 6, but stronger in adolescents and adults.
- Shared decision making concerning the objectives on the basis of the diagnostics and in consultation with the child who stutters and the parents is warranted given the multifactorial nature of stuttering.

5. Effectiveness of Stuttering Therapies in Adolescents and Adults

The effectiveness of various stuttering therapies for adolescents and adults will be described in Chapter 7.

Recommendations

13. The treatment of stuttering for adolescents and adults should be individualized. Therapy may take place in an individual and/or group setting. The treatment plan is established in a dialogue between the Speech and Language Therapist (SLT) and the Person Who Stutters (PWS) (' shared decision making '); the treatment plan contains all ICF-elements. The wishes and needs of the PWS form the basis of that treatment plan.
14. Depending on what has been agreed by the person who stutters and the speech & language therapist/fluency specialist, the treatment will focus on the psychosocial aspects (emotional; and cognitive reactions to speaking), on verbal-motor aspects or on both. Cognitive behavioural therapy is recommended for the treatment of psychosocial aspects.
15. Other elements of stuttering therapy should include: *Promoting transfer* - it is essential that the PWS applies the skills learned and insights in daily living. *Promoting and maintaining self-management* - the PWS is able to evaluate the stuttering and the associated behaviour and adjust this if necessary.
16. In general, the use of Altered Auditory Feedback based devices (AAF) is not recommended. However, in specific circumstances, such equipment may reduce the stuttering rate in some PWS. One should note that this effect may not be maintained.

Rationale for the recommendations

- Various stuttering therapies have positive effects directly after treatment on the stuttering frequency; these, however, vary in impact. Some studies report that positive effects are maintained six months after therapy, yet others report (some) recurrence. The certainty around the effect size is limited for the stuttering frequency outcome measure, through limitations in study design and sample size. Outcome measures other than stuttering frequency, such as avoidance behaviour (situation and word avoidance),

naturalness of speech, participation or quality of life have not been sufficiently investigated.

- The scientific literature does not provide any strong evidence that one stuttering therapy is (much) more effective than another.
- It is therefore appropriate to jointly determine the choice of a specific therapy based on the wishes and needs of the person who stutters.
- The working group, in drafting these recommendations, has been unable to attach any conclusive figures on the costs of stuttering therapy, due to the lack of information about the current costs.
- The recommendation for therapy in a group setting rests on the experience of members of the working group and the experiences of persons who stutter, as expressed in the context of a focus group organised as part of this guideline. Therapy in a group setting may enhance the motivation for therapy and be a supplement to peer-group contact.

6. Effectiveness of Pharmacotherapy for Adolescents and Adults

Chapter 8 discusses the effectiveness of various pharmaceuticals for adolescents and adults who stutter. There will only be a brief description of the current state of affairs given that pharmacotherapeutics are not applied in the Netherlands. Any evaluation in respect to the certainty of the effect size is therefore also absent.

Recommendation

17. Use of pharmaceuticals in the context of stuttering therapy is not recommended. Where there is co-morbidity and stuttering, it is recommended that an appropriate choice and dose of pharmaceuticals is sought in consultation with the PWS (and their direct environment) and the prescriber.

Rationale for the recommendations
- Virtually all pharmaceuticals are associated with side-effects after long-term use. Given the absence of significant effects of pharmaceuticals on the one hand and potential side-effects on the other, there is no reason to recommend the (long-term) use of pharmaceuticals.

7. When and for what Reasons should a Patient who Stutters be Referred by a Speech & Language Therapist to a Fluency Specialist or another Healthcare Provider?

The treatment of stuttering can be complex and will not always only be carried out by a speech therapist or fluency specialist. Chapter 9 describes what the indications are to refer to the fluency specialist as well to a different healthcare provider. This document provides more clarity about when a referral should be made and to which healthcare provider.

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Recommendations

Expertise

18.	The speech & language therapist needs to be able to implement the recommendations in this guideline regarding diagnostic and therapeutic approaches. If the SLT lacks the experience or knowledge to do so, the client should be referred to an SLT with more expertise in the field of stuttering or to a Fluency Specialist.

Social/emotional problems

19.	Where there is a suspicion of social anxiety or depression in the person who stutters based on findings from the Behaviour Assessment Battery (BAB), the speech & language therapist or fluency specialist needs to consolidate this with the Strength and Difficulties Questionnaire or the Four Dimensional Symptoms List. Following a positive indication the speech & language therapist or fluency specialist should consult with the PWS or the parents about a referral to the GP for a potential referral to a psychologist or psychiatrist.
20.	In case of stuttering children and young people, the SLT should explore the problem of bullying. This may include a suitable questionnaire for bullying. If bullying is identified, the SLT should discuss this with the parents and agree a plan of action.

Course of therapy

21.	During the assessment, the SLT will provide information to the PWS regarding the treatment options in order to enable an informed choice regarding the treatment. The SLT should provide information regarding other relevant resources and websites.
22.	If a child who stutters up to aged 6 years has not made progress with 11 to 12 therapy sessions or within 3 months, a Fluency Specialist should be consulted.
23.	If during two-or three-monthly evaluations of the therapy process, the realistic and achievable treatment goals may have not been sufficiently realized (according to the PWS or to the SLT) the SLT should discuss the possibility of referral to another practitioner.

24.	The SLT and other professionals involved with PWS in a specific region set up a working agreement concerning collaboration, referring and information transfer. When referring to another SLT or Fluency Specialist, the SLT formulates specific questions incorporating all relevant ICF elements.

Rationale for the recommendations

- The complexity of stuttering and the associated social/emotional problems requires specific skills from the therapist.
- Continuing with a therapy that is having an inadequate effect is undesirable.
- In the treatment of young children aged up to six who stutter a significant improvement in stuttering occurs on average after 11-12 treatment sessions (Kingston, Huber, Onslow, Jones & Packman, 2003; Millard, Nicholas & Cook, 2008; Yaruss, Coleman & Hammer, 2006).
- Some PWS develop serious emotional problems, such as depression or social anxiety (Blumgart, Tran & Craig, 2010; Iverach & Rapee, 2013; Koedoot, Bouwmans, Franken & Stolk, 2011; Tran, Blumgart & Craig, 2011).
- Children and adolescents who stutter have a greater chance of being bullied and of experiencing negative reactions from their peers (Blood et al., 2010; Langevin, 2009).
- The fluency specialist has additional expertise in treating stuttering.
- The speech & language therapist or fluency specialist are not equipped to deal with complex or severe social/emotional problems irrespective of whether this is caused by stuttering.
- The Strength and Difficulties Questionnaire (SDQ) is recommended for screening for social/emotional problems in children aged 3 to 13 (Ruiter & Jong, 2013). This is a simple tool to use and is freely available (www.sdqinfo.org).
- The Four Dimensional Symptoms List (4DSL) or Depression Recognition Scale (DRS) are recommended for screening for social/emotional problems in adolescents and adults (Ruiter & Jong, 2010).

8 How should Proper Aftercare be Organised and Implemented?

In Chapter 10 adequate aftercare is defined and the organisation of aftercare for people who stutter is described. In the context of this guideline the working group understands aftercare as: the pathway following the end of therapy with the associated care and aftercare appointments.

Recommendation

25. Any therapeutic approach to stuttering must include methods which promote long-term effects, and deal with possible setbacks. As a part of the therapy process, the way in which follow up support is organized, is defined by the SLT and the PWS in collaboration with his environment. At the end of the clinical treatment the SLT proposes a programmed approach for an individualized follow up program lasting for 2 years. In order to prevent relapse and to promote long-term effects, the SLT suggests the PWS and his environment to contact other PWS, e.g. activities organized by patients' associations

Rationale for the recommendations

- Recurrence occurs frequently, and significant benefits may only be achieved if the PWS has internalised the transfer of therapy into non-clinical situations (i.e. during participation in society) and has an aftercare trajectory at hand.
- The concepts aftercare and self-help are not always clearly distinguished from each other in the literature around stuttering.
- The aims of good quality aftercare need to be defined.
- In the literature on stuttering and in clinical practice the usual, desirable duration of aftercare is two years.
- The working group was unable to establish any accurate estimates on the costs of aftercare in drafting these recommendations, mainly through the lack of information about current costs.

Chapter 1: General Introduction

1.1 Background and Motivation

The worldwide life time incidence of stuttering ranges from 5 to 8 %: calculated for children, values up to 17% are reported. Stuttering persists (i.e. the prevalence) in approximately 1% of the population. Stuttering may lead to social and relational limitations. The treatment and/or coaching of people who stutter and their environment may provide an important contribution to reducing these limitations.

Various techniques based on different therapeutic formats are used to treat stuttering. The success of the therapy provided varies. It is not always clear to the speech and language therapist, fluency specialist, person who stutters or the healthcare insurer as to which form of therapy is best for a given situation.

Until now, there has been an absence of evidence-based guidelines on stuttering using the recently adopted GRADE procedure (Guyatt, 2008).

Evidence-based guidelines are "documents with recommendations, aimed at improving the quality of care, based on systematic review of scientific research and deliberations about the advantages and disadvantages of the different care options, supplemented with expertise and experiences of care professionals and healthcare users." (Guideline for Guidelines, 2011). Guidelines aim to :

- make the rapidly growing information influx more manageable;
- reduce the undesirable variations in treatment between healthcare providers;
- base clinical treatment on scientific evidence rather than on experience and opinion;
- provide referrers and patients, among others, more transparency

Guidelines are a means to make the best care explicit on the basis of three sources: scientific evidence, the expertise of professionals in the relevant fields and the expertise of patients. Guidelines also form an important basis for new clinical and practice-oriented scientific research initiatives, given that the aforementioned literature reviews exposed gaps.

There are three English language guidelines available in the field of stuttering. These guidelines are out of date and are not based on the GRADE criteria. The most influential guidelines are those that have been developed at a national level and are being interpreted at a local level (Barkham et al., 2010). The Dutch Association for Logopedics and Phoniatrics (NVLF), the Dutch Association for Stuttering Therapy (NVST) and Demosthenes, the patient

association for (parents of) children and adolescents suffering from stuttering have, therefore, expressed the wish to define how the diagnostics, treatment and follow-up care of clients who stutter should be designed. Demosthenes has also emphasised that there has to be information available to people who stutter to enable them to primarily engage with a speech and language therapist/fluency specialist. The CBO was approached by the NVLF, NVST and Demosthenes for their great experience in developing guidelines. The CBO is intent on improving patient care in collaboration with professionals, patients and healthcare organisations and wishes to contribute to the improvement of the care of people who stutter. Demosthenes, the NVLF, NVST and the CBO have jointly drawn-up a plan to develop the Guideline on Stuttering, opting for the method of guideline development tailored to the requirements as set for evidence-based guideline development internationally.

The presentation of the writing process for this guideline and several examples from it during the 8th World Congress on Fluency Disorders, 6– 8 July, 2015 Lisbon, Portugal, aroused considerable enthusiasm among the audience and current authors were asked to provide this guideline in English. This translation has been duly performed by Univertaal. During that process, some remaining administrative errors or ambiguities have been clarified collectively by three people from the whole group of authors, i.e. LCO, MAJP and EJEGB. Further, we would like to thank Elaine Kelman for technical linguistic advices. Recent data as to early intervention has been published in the meantime and these references were included. Thereby, some sentences in Chapter 5 could be strengthened. The entire English translation has been offered to the whole group of original authors, as well as to the sponsors. This text will also be offered to the International Stuttering Association and to the International Fluency Association for comment. Such comments will be gladly received by MAJP to possibly improve the text. Whereas the text is based on international publication, it has not been the aim to provide a guideline which could be used globally on a specific country-wise level. Rather, national bodies may depart from the text after approval from the ISA and IFA mentioned, in order to adopt the text to specific national circumstances – like insurance systems and availability of certain modes of therapy. Guidelines are subject to specific national agreements; they do not pretend to give a rule box, but rather a tool box. It remains the individual responsibility of the clinician to utilise this guideline. Current authors decline such a responsibility.

1.2 Aims of the Guideline

The guideline is aimed at improving the quality and effectiveness of the care provided by speech and language therapists and fluency specialists to people who stutter and their

environment. This guideline has clear recommendations in respect of optimal care to people who stutter in terms of diagnostics, treatment, referral and follow-up care taking into consideration the current state of scientific literature and insights within professional practice during 2013. Furthermore, the guideline is aimed at enhancing knowledge about stuttering within speech and language therapists, fluency specialists, doctors, educational establishments, people who stutter and their parents.

1.3 Target Population

The guideline provides recommendations about diagnostics, treatment, follow-up care and management of children, adolescents and adults who stutter.

Developmental stuttering begins, in general, before age six, usually between two and a half and four years of age. The most common form of stuttering is referred to as developmental stuttering. This may be contrasted with other forms of stuttering which are due, for instance, to a neurological condition, a trauma or through emotional stress.

This guideline is exclusively aimed at developmental stuttering and the term 'stuttering' applies in general to this phenomenon amongst speech and language therapists and fluency specialists.

1.4 Intended Users

The guideline was drawn-up for speech and language therapists and fluency specialists. Other healthcare professionals coming into contact with adults, children and adolescents who stutter, such as general practitioners, adolescent and paediatricians, ENT physicians, psychologists and occupational health physicians may consult the guidelines in order to optimise care to people who stutter. Furthermore, the guideline may also be used by healthcare insurers and people who stutter in order to enable them to know what they may expect in terms of the care provided.

1.5 Principal Parties Involved in the Guideline Development

The initiative for developing and implementing the evidence-based clinical Guideline on Stuttering in children and adults was undertaken by the Dutch Association for Stuttering

Therapy (NVST), Demosthenes, the patient association for (parents of) children and adults who stutter and the Dutch Association for Logopedics and Phoniatrics (NVLF). The guideline was developed under the auspices of the NVLF; the NVLF is the sponsor and patron of the guideline.

1.6 Working Group and Advisory Panel

A working group was established for the development of this guideline in 2012. This working group comprises representatives from the most relevant professional organisations involved in the diagnosis and treatment of stuttering, as well as representatives from the patients' association and guideline methodologists from the CBO.

The working group was responsible for outlining the draft guideline and determining the definitive guideline text. In constructing the working group, attention was paid to the geographical spread of the working group members as well as ensuring a proportional representation of the various associations and academic backgrounds. The members acted independently and were mandated by their association to participate in the working group. All members of the working group completed a declaration of interests. None of them reported any relevant competing interests.

A number of professional organisations indirectly involved in the diagnostics and treatment of adults and children who stutter participated in an advisory panel. They were asked to detail which important bottlenecks they experienced in the care of children and adults who stutter. During the comments phase, members of the advisory panel were asked to provide feedback on the draft guideline.

1.7 Patient Perspective

The patient perspective took a central role in the drafting of this guideline. Patient representatives from Demosthenes (patient association) participated in the working group. A focus group was also formed with people and parents of children who stutter, irrespective of whether they were members of the patient association Demosthenes or not; this enabled the population of people who stutter in the Netherlands to be represented as evenly as possible in drafting the guideline.

1.8 Method

a. Issues and Principal Questions

In order to depict the issues facing people who stutter, they and the associated healthcare professionals were invited via various channels to complete a questionnaire about those problems. An analysis of these issues was completed from both the patient and healthcare professionals' perspective based on the results of the questionnaire. The issues were explored in further depth in a focus group meeting with people who stutter and parents of children who stutter. An outline of the principal questions was formulated on the basis of the analysis of these issues; this was subsequently evaluated by the working group. Once the comments from the working group had been incorporated, a definitive list with the principal questions was devised; this focused on the most important issues in daily practice (Appendix 7). The principal questions form the basis of the different chapters in this guideline.

b. Working Group Approach

The working group dedicated approximately a year and a half to answering the principal questions and preparing the text for the draft guideline. The methodologists provided the descriptions for the scientific evidence. The conclusions from the literature formed the basis for drafting the recommendations. The designated working group members produced a presentation which was discussed during a meeting of the group and supplemented with practical information as required. The recommendations were created on the basis of (informal) consensus within the working group. For a limited number of the principal questions the working group members wrote a text that was not based on the results of the systematic literature review, as it was clear in advance that there was no, or barely any, scientific evidence available for these areas. The texts were discussed during plenary meetings and approved once the comments had been incorporated. The draft guideline approved by the working group was subsequently offered to the professional and patient associations involved for comment. A definitive draft of the guideline was approved on 8 October 2014 by the working group once these comments had been incorporated, and sent for authorisation to the relevant professional organisations. The guideline was approved at the end of October 2014 by the boards of the professional associations.

c. Scientific Evidence

The guideline is based on evidence from published scientific research. Relevant databases such as the Cochrane Library, MEDLINE, EMBASE, PsycINFO and CINAHL were systematically searched for relevant articles. Each principal question had a separate search strategy; these have been described in brief for each principal question and may be requested from the CBO.

In addition to the literature from the search, publications were also included for a number of questions from the archives belonging to working group members, provided they satisfied the inclusion criteria. Those forms of therapy and treatment publications for which there were no publications that satisfied the inclusion criteria were not used for the scientific evidence, but were potentially discussed in the context of the professional perspective as part of the other deliberations.

Following the selection of the most relevant literature, the articles relating to therapeutic interventions were assessed for the quality of the research and graded according to the degree of evidence. Those studies involving the validity, reliability and standardisation of diagnostic instruments were not graded according to the quality of evidence.

The quality of evidence - also designated as the level of certainty of the effect size for an outcome measure - was assessed using GRADE (Guyatt, 2008). GRADE is a method that allocates a grade to the quality of evidence according to the outcome measure in an intervention based on the confidence in the estimation of the effect size (Tables 1 and 2).

Table 1. Categorisation of the quality of evidence or degree of certainty in respect of the effect size for an outcome measure according to GRADE (Balshem et al, 2011)

Degree of certainty of the effect size	Description
High	The true effect lies close to that of the estimate of the effect.
Moderate	The true effect is likely to be close to the estimate of the effect, but there is a possibility that it is substantially different
Low	The true effect may be substantially different from the estimate of the effect.
Very low	The true effect is likely to be substantially different from the estimate of the effect.

Table 2. The quality of evidence or degree of certainty in respect to the effect size is determined on the basis of the following criteria (Balshem et al, 2011)

Type of evidence	RCT starts in the "high" category.		
	Observational study starts in the "low" category.		
	All other study types start in the "very low" category.		
Negative evaluation	Risk of bias	1	Serious
		2	Very serious
	Inconsistency	1	Serious
		2	Very serious
	Indirect evidence	1	Serious
		2	Very serious
	Inaccuracy	1	Serious
		2	Very serious
	Publication bias	1	Likely
		2	Very likely
Positive evaluation	Large effect size	1	Large
		2	Very large
	Dose-response relationship	1	Evidence of a gradient
	All plausible confounders	1	Would reduce a demonstrated effect
		1	Would suggest a spurious if no effect was observed

d. Development of the Recommendations

Other aspects were of importance in addition to the scientific evidence in producing the recommendations, for instance: balance of desirable and undesirable effects, burden of therapy, patient preference, professional perspective, availability of special techniques or expertise, organisational aspects, and social consequences or costs.

International guidelines relating to the diagnosis, treatment, management and assessment of stuttering were consulted in the process of detailing the professional perspective. Databases

from the US National Guideline Clearinghouse (www.guideline.gov) and the Guidelines International Network (www.g-i-n.net) were consulted for this.

These aspects are discussed after the "Conclusion" under the heading "From Evidence to Recommendations". The recommendations which were ultimately formulated are the result of the available evidence in combination with these other deliberations. This procedure and the format of the guideline itself were specifically followed in order to enhance its transparency. Meanwhile, allocating a rationale to the recommendations is an attempt to bring this transparency into effect. This provided space for an efficient discussion during the working group meetings, and furthermore increased clarity for the user of the guideline.

1.9 Dissemination and Implementation

The implementation of the guideline and the practical feasibility of the recommendations were envisaged in all the different phases of its development, with explicit concern for any factors that could promote or hinder its implementation in practice. A patient version of the guideline was also developed. The guideline is being disseminated across all the relevant professional groups, patient organisations and educational establishments. Moreover, it is being publicised via publications in journals and websites of the various organisations. The guideline may also be downloaded from:

http://nvlf.logopedie.nl/site/inhoudelijke_richtlijnen

1.10 Legal Significance of the Guidelines

The guidelines are not statutory stipulations; they are wide-ranging evidence-based insights and recommendations for good quality care. Given that the guidelines are based on the "average patient", healthcare providers may, in individual cases, deviate where necessary from the recommendations in the guideline. Deviations from the guidelines - if the patient's situation requires this - may sometimes even be a necessity. Where there has to be a deviation from the guideline this should be supported by evidence, documented, and, where necessary, undertaken in consultation with the patient.

1.11 Review of the Guideline

After consultation with the associations participating in the guideline, the NVLF and NVST will determine no later than 2019 whether this guideline is still up-to-date. If needed, a new working group will be created to review (parts of) the guideline. The validity of the current guideline will expire should new developments be reason for an update or review.

1.12 Financing

The guideline was made possible partly through co-financing from the Dutch Association for Logopedics and Phoniatrics, Demosthenes, the patient association for (parents of) children and adults who stutter, the Dutch Association for Stuttering Therapy (NVST) and the Damsté-Terpstra Fund.

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Chapter 2: Stuttering and Speech & Language Therapy

2.1 Definition of Stuttering

Stuttering is a disorder in the fluency of speech. Characteristics of stuttering include the involuntary repetition and prolongation of sounds and syllables. There may also be tense pauses or blocks disrupting the rhythm of speech. The repetitions, prolongations and blocks are designated with the term "core stuttering behaviour" or "core stuttering".

In addition, there is also "secondary stuttering behaviour" which occurs as a response to the core stuttering behaviour. Examples are "flight behaviours", varying from choosing synonyms for the feared words to opting for social isolation to avoid speaking and concealing the stuttering out of anxiety, frustration and shame. There are also "fight behaviours" varying from unnatural eyeblinking to involuntary movement of the extremities. Stuttering may therefore have significant consequences to the personal and social functioning of the PWS.

The level of stuttering varies according to the situation. Stuttering may be exacerbated in speaking situations creating anxiety, such as speaking in front of a group or talking on the telephone. Speaking to a small child or talking when you are alone are examples of situations were speech becomes more fluent. Singing is also almost always fluent.

Stuttering arises, in general, before age six, usually between two and a half and four years of age. The most common form of stuttering is referred to as developmental stuttering. This may be contrasted with other forms of stuttering which, for instance, arise as a result of a neurological condition, a trauma or through emotional stress.

This guideline is exclusively aimed at developmental stuttering. Any reference throughout this document to "stuttering" is intended to refer to "developmental stuttering", as this is the term generally used amongst speech and language therapists and fluency specialists.

2.1.1 Epidemiology

The prevalence of stuttering, the number of people who stutter at a specific moment, is estimated at 0.72% to 1% of the world population. As stuttering is often remedied in children under the age of six, the prevalence in this young population group is greater than in an older population. The life time incidence of stuttering (new cases per year) has been fluctuated around 5%, whereas in the last decade higher values have been reported - around 8%. When measured in children, even much higher values have been reported, up to 17%.

The details about the prevalence and incidence differ per study. This is due, amongst other things, to the differences in the definition of stuttering, the age of the study groups and the research methods. The recovery percentages described are not universal either. The

percentage recovery of stuttering is estimated at 50% to 90% (spontaneous or with the help of therapy) (Yairi & Ambrose, 2013).[1] Shortly after the first stuttering symptoms have been observed the chance of spontaneous recovery is estimated at approximately 75%. The chance of spontaneous recovery decreases in proportion to the amount of time the stutter has been present. Most recovery takes place in the period before the 7th year of life, but this may continue throughout the teenage years. Recovery of stuttering remains possible in adolescents and adults, but is relatively rare (Finn et al., 2005; Kell et al., 2009).

Stuttering is more common in men than in women. There are more boys who start stuttering at a young age than girls (2:1) and recovery in girls is, in addition, greater than for boys. The ratio of men to women who stutter in adults is approximately 4:1 (Yairi & Ambrose, 2013). Stuttering occurs across the entire world, in each race and in each culture.

2.1.2 Cause of Stuttering

There are numerous theories about stuttering, however "the" cause is as yet unknown. Stuttering is viewed as a multifactorial disorder: multiple factors are of influence in the occurrence of stuttering. The most important factors are discussed below.

Genetic factor

Twin studies and family studies have shown clear evidence of the existence of a strong genetic factor (Bloodstein & Bernstein Ratner, 2008; Rautakoski et al., 2012). It has been calculated that approximately 80% of stuttering may be explained due to the genetic background. Evidence has been found that various genes and risk alleles could play a potential role in stuttering (Raza et al., 2012).

Brain structure and functions

Since 1920, research has been undertaken into the functional and structural differences in the brains of people who stutter. Electrophysiological studies and fMRI studies have shown that there is more right hemisphere activity during speech tasks, in particular an overactivation of the right pre-central sensorimotor cortex (e.g. De Nil, Kroll & Houle, 2001). Underactivation has been found in areas associated with auditory functions. This potentially means that mechanisms associated with hearing your own speech are not functioning well (Brown et al., 2005). In addition to this there is evidence that the basal ganglia have a role in stuttering (Alm,

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[1] Yairi and Ambrose have suggested that the studies investigated did not always make a distinction between spontaneous recovery or recovery due to treatment. The estimate for spontaneous recovery is therefore significantly lower than 94%

2004).[2] Anatomical differences have also been found in brain structure, particularly in the grey and white matter (Chang et al., 2008). These differences are also found in children who stutter. These are a few examples of results from brain studies that highlight the presence of a neurological factor. The question as to whether the differences uncovered in the brain is a cause or a result of stuttering is still open for discussion. Furthermore it is not clear which of the differences found point to the pathology.

Sensorimotor system

Various research into the sensorimotor systems of PWS has shown that it reacts more slowly in PWS than in control groups, and that it is less stable. This applies both to tasks that are related to speech (including fluent speech), as well as other sensorimotor skills, such as rhythmic handclapping tasks (incl. Smith et al., 2010; Oleander, Smith & Zelaznik, 2010).

Language factor

Stuttering occurs in the period in which language is developing, at a point when children are starting to use longer sentences and are experiencing a large expansion in vocabulary. It is apparent from the results of various studies into the relationship between language skills and stuttering that subtle differences have been found in the language skills of children who stutter compared to those who do not stutter, both in receptive and in productive language (Bloodstein & Bernstein Ratner 2008; Ntourou et al 2013; Yairi & Ambrose, 2005; Seery & Yairi, 2013; Anderson et al, Reilly, 2013). These differences are not evident in standardised language tests. Nearly all studies have shown that children who stutter have more phonological problems than children with fluent speech. Language factors may, in addition, have an effect on the stuttering moment (at the start of the sentence, using a content word, particularly in complex linguistic structures). There have been no consistent indications that stuttering is more prevalent in multilingual than monolingual children (Shenker, 2011).

Personality

A lot of research has been undertaken into the personality structure of PWS, however there is no evidence of an underlying neurotic or personality disorder as a cause of stuttering. The emotional problems experienced by the PWS are a reaction to rather than a cause of the stuttering and are therefore secondary. Research into temperament of children who stutter has investigated certain temperamental characteristics that could be associated with stuttering. Different studies have shown conflicting results which means it is difficult to draw clear conclusions. A number of studies have shown that children who stutter are less

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[2] The basal ganglia (or basal nuclei) are a group of nuclei of diverse origin in the brains of vertebrates, which operate as a collective functional unit and are associated with, amongst others, routine behaviours, as well as cognitive and emotional functions.

adaptable, have a shorter attention span and a higher amount negative emotions (Eggers, De Nil & Van den Bergh, 2010; Kefalianos et al., 2012). The question of whether this is a cause or perhaps a consequence of stuttering cannot be answered at this moment.

Environment

Research into the difference in the environment of children who speak fluently and those who stutter has not provided any clear results. Parents of children who stutter do not appear to differ in terms of character or parenting style. A stressful environment or event is, however, viewed as a potential factor that may evoke or maintain stuttering.

Learning factors

Classical and operant conditioning processes ensure that stuttering continues to develop further. The secondary behaviour associated with stuttering may be explained through a learning theory perspective. For instance, certain situations may be associated through previous learning with stuttering such as making a telephone call (classical conditioning). An example of operant conditioning is struggling on through a stuttering moment, enabling someone to continue talking: the utilisation of motor effort is rewarded.

2.1.3 Stuttering and the International Classification of Functioning Disability and Health (ICF) Model

According to the International Classification of Functioning, Disability and Health (ICF) model health problems are categorised in terms of anatomical features, functional disorders, limitations in activities and problems in participation. Whether a person experiences problems in activities and participation is not solely dependent on the presence and severity of the functional disorder; it also depends on personal and external factors. These are consequently a part of the ICF model.
Figure 1 provides the graphical representation by Yaruss and Quesal (2004) of how the health problems associated with stuttering and the factors that may influence these problems can be categorised according to the ICF terminology.
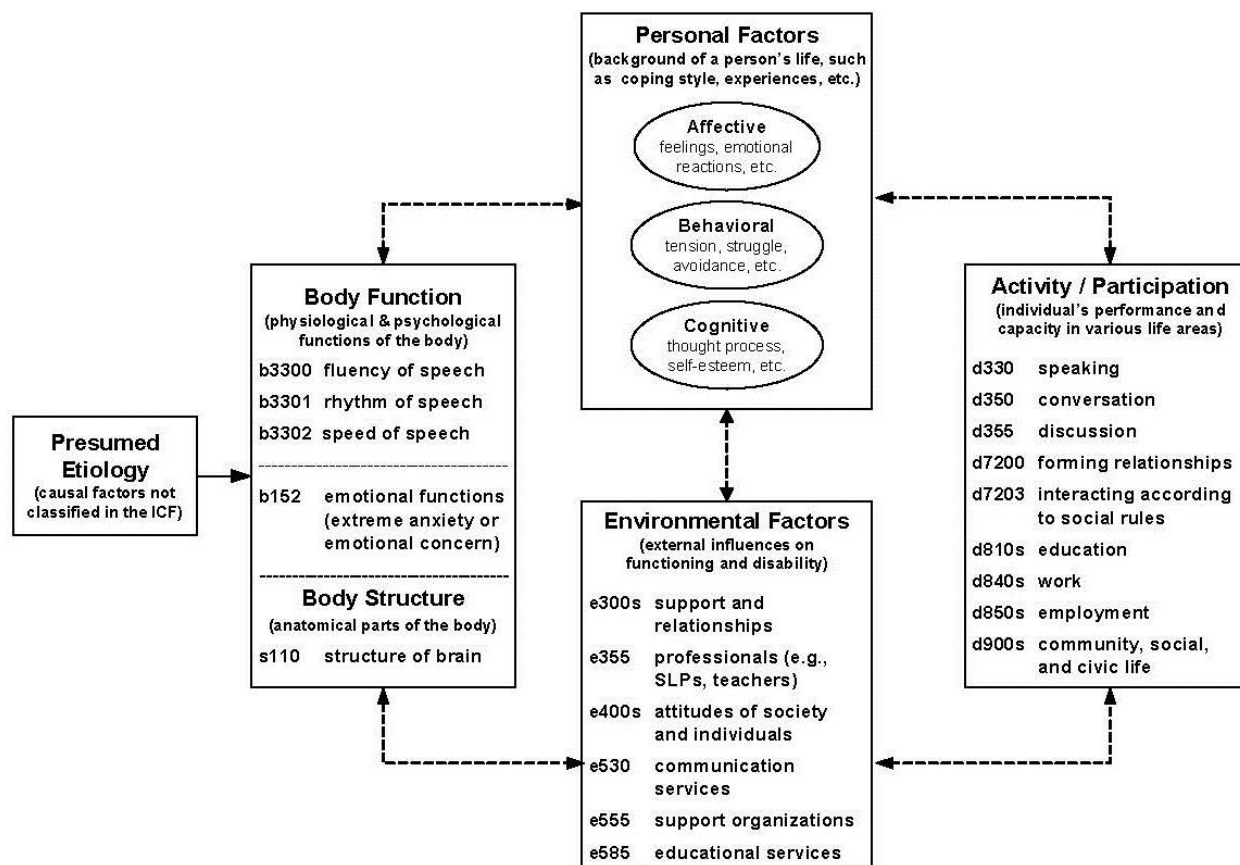
Figure 1. Graphical representation of how the World Health Organization's International Classification of Functioning, Disability, and Health (ICF) can be applied to the stuttering disorder (Yaruss & Quesal, 2004).

2.1.4 Development of Stuttering

Many children experience a period during speech / language development between the ages of 2 to 5, where speech becomes dysfluent. Characteristics of these normal dysfluencies are, for instance, easy repetitions of words or part of a sentence, interjections and revisions. There are no secondary behaviours and the children usually do not notice the dysfluency.

These normal dysfluencies also occur in young children who stutter. In addition to these the children will have a different type of stutter-like dysfluency and the frequency of the dysfluencies will be higher than normal. The development of stuttering differs between individuals, however stuttering behaviour does demonstrate common characteristics for each age group. The evolution of normal dysfluency into confirmed stuttering is represented in Table 1. This has been described in five phases. The hierarchy sketched out in Table 1 is often experienced as a sequence by the child, however it may also very well be that the child displays normal dysfluency one day and the features of intermediate stuttering the next. Some children develop immediately to the phase of beginning stuttering. There are therefore different developmental pathways.

Development level	Core behaviours	Secondary behaviours	Feelings and attitudes
Normal dysfluency	10 or fewer dysfluencies per 100 words; one-unit repetitions; mostly repetitions, interjections and revisions	None	Not aware, no concern
Borderline stuttering	11 or more dysfluencies per 100 words; more than two units in repetitions; more repetitions and prolongations than revisions and interjections	None	Generally not aware; may occasionally show momentary surprise or mild frustration
Beginning stuttering	Rapid, irregular and tense repetitions may have fixed articulatory posture in blocks	Escape behaviours, such as eye blinks, increases in pitch, or loudness as dysfluency progresses	Aware of dysfluency, may express frustration
Intermediate stuttering	Blocks in which sound and airflow are shut off	Escape and avoidance behaviours	Fear, frustration, embarrassment and shame
Advanced stuttering	Long, tense blocks; some with tremor	Escape and avoidance behaviours	Fear, frustration, embarrassment, and shame; negative self-concept

Table 1. Characteristics of five development levels of stuttering (after Guitar, 2014, p. 132)

2.1.5 Diagnosis

During diagnosis, stuttering will be investigated as thoroughly as possible, with an assessment on all of the aspects described in the ICF schedule (Figure 1). In young children with fluency problems there is an important question as to whether it is a case of a normal dysfluency period or an early form of stuttering. The information provided by the parents is important specifically because stuttering may still vary significantly during the early phases. Developmental stuttering should be distinguished from other forms of non-fluent speech. If stuttering suddenly arises or arises after age 7 then this is viewed as a warning sign for a potentially different form of stuttering. In addition, there are non-fluency disorders that may be confused with stuttering. The most important of these are explained below.

Cluttering

Cluttering is a form of non-fluent speech where the speaker is not sufficiently able to adapt to the demands of the speech motor system and/or linguistic demands at that moment in time (van Zaalen, 2009). Cluttering is characterised by a rapid or irregular speech rate, reduced intelligibility and a greater than normal number of dysfluencies, the majority of which is

atypical for stuttering. Cluttering and stuttering often occur together, but they are classified as separate disorders in the International Classification of Functioning, Disability and Health. Experts have difficulty arriving at an agreement when a diagnosis of cluttering has to be determined, partly as the characteristics of cluttering often co-occur with stuttering and pure cluttering is rare.

Neurogenic stuttering

Neurogenic stuttering is an acquired speech disorder characterised by stuttering-like dysfluencies following brain damage. It may arise after a Cerebral Vascular Accident (CVA), as well as after brain trauma or as a result of a degenerative neurological condition. This form of stuttering occurs particularly in adults. In approximately 50% of cases there is co-morbidity with aphasia or dysarthria, occasionally also verbal apraxia (Theys et al., 2012). The moment of onset and the patient's medical history are of importance for the differential diagnosis with developmental stuttering. A typical feature of neurogenic stuttering is that stuttering moments may occur at any location in a word, and not just at the initial sounds or syllables notable in stuttering.

Psychogenic stuttering

Psychogenic stuttering is rare and may arise after a traumatic experience, after a long period of stress or in combination with a psychiatric disorder. The medical history is of importance for the differential diagnosis. A neurogenic cause needs to be excluded.

Pharmacogenic stuttering

Some medicines, such as selective serotonin re-uptake inhibitors or tricyclic antidepressants may give rise to speech dysfluencies as a side effect in patients. (Krishnakanth, Haridas Phutane, & Mularidharan, 2008).

2.2 Treatment

This chapter describing treatment methods provides an overview of stuttering therapy that is currently applied in the Netherlands.

2.2.1 Healthcare Professionals

Therapy for stuttering is provided by speech and language therapists and by speech & language - fluency specialists. Fluency specialists are speech and language therapists who having completed an undergraduate degree in speech & language therapy have pursued an

NVST (Dutch Association for Stuttering Therapy) approved additional training programme, or who have specialised in stuttering therapy after completing a number of post-vocational courses. Speech & language therapists and fluency specialists need to satisfy the requirements of the Paramedical Quality Registry, which stipulates quality standards for work experience and promoting expertise. There are additional quality standards for speech & language - fluency specialists who are registered with the NVST.

The Netherlands has seen a range of widely varying stuttering programmes offered by providers other than speech & language therapists or fluency specialists. Providers of such, usually commercial, stuttering programmes have a diverse background in respect to their training, sometimes using their personal experience rather than a standardised approach, and do not have to satisfy the quality standards applied to speech & language therapists and fluency specialists. A shared issue among these programmes is the lack of evaluable data in scientific litterature. The treatment as described below is based on scientific literature and the current approach adopted by speech & language therapists and fluency specialists.

2.2.2 Approach

The approach taken by the speech & language therapist and fluency specialist is methodical and contains certain phases and steps (see Figure 2). It is a cyclic process: where necessary previous steps may be re-visited.

Phase 1 Speech & Language Diagnostics
step 1 intake/screening
step 2 case history
step 3 speech & language assessment
step 4 analysis (including formulating a diagnosis)

Phase 2 Treatment
step 1 treatment plan
step 2 treatment
step 3 evaluation
step 4 completion
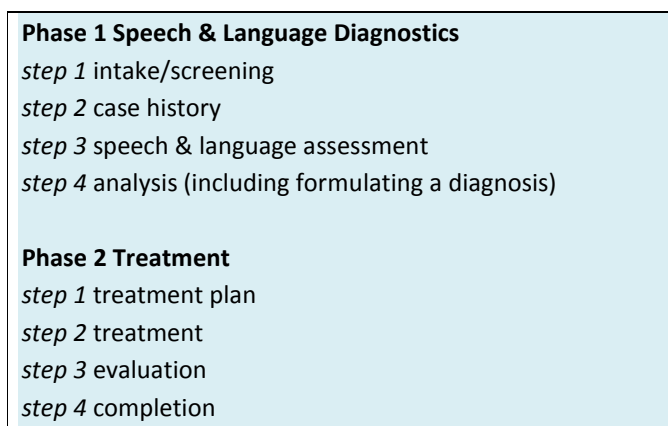
Figure 2. Methodical steps for speech & language therapy (NVLF, 2013)

All aspects of the stuttering, as described in the ICF schedule (see Figure 1), are detailed as thoroughly as possible during the diagnostic phase. An individual treatment plan is drawn-up in consultation with the PWS on the basis of the presenting problem and the outcomes of the diagnostic assessment. Reducing or changing core stuttering behaviour and the (observable) secondary stuttering behaviour is often an important treatment aim; however reducing anxiety and utilising cognitive therapy are essential components of stuttering therapy for many PWS that are offered by speech & language therapists and fluency specialists. Therapy

for young children will include parents and the environment. The treatment aims will be regularly evaluated during therapy and amended as required.

The fluency specialist will work alongside other healthcare professionals and will refer the patient to others if required due to the nature or complexity of the problem. For instance, a speech & language therapist may refer the patient to a specialist fluency therapist, however there will also be collaborations with psychologists, social workers, general practitioners, paediatricians, ENT physicians and academics. The speech & language therapist provides a report about the treatment to the general practitioner.

2.2.3 Treatment of Adults and Adolescents

Therapeutic methods and techniques

There are two distinct principal approaches within the different therapeutic methods: stuttering modification therapy and fluency shaping therapy.

The primary central aim of stuttering modification therapies is reducing anxiety (desensitisation). The PWS is taught to stutter freely and easily, and how to properly deal with dysfluent speech. This is referred to as easy or relaxed stuttering.

Fluency shaping therapies teach the PWS primarily to have controlled fluent speech using speech techniques; subsequently they are taught how to apply this technique in different speaking situations.

Various therapeutic methods and techniques are deployed with these two principal approaches. Examples of therapeutic methods for anxiety reduction and behaviour modification include systematic desensitisation, gradual confrontation, relaxation and cognitive training. Social skills training may also be a part of therapy.

Various techniques are utilised for learning fluent speech such as prolonged speech, easy onsets, rhythmic speech, reduction of speech rate and breathing techniques.

In general, speech & language therapists and fluency specialists in the Netherlands use an integrated approach, where aspects of both stuttering modification, as well as fluency shaping methods are used depending on the needs of the individual client. Therapy is offered both at an individual and group level. A follow-up trajectory is usually provided as a recurrence may arise after therapy.

Devices

Various devices are available on the market that may be used to promote fluent speech. The principle of these devices is based on a modified feedback of an individual's speech, for instance by masking it with a sound or by feeding back the individual's speech at a delayed rate or changing it via a hearing aid (e.g. Delayed Auditory Feedback (DAF); Frequency Altered Feedback (FAF)). There are also devices available to support rhythmic speech.

Pharmacotherapy

Pharmacotherapy is not provided for stuttering in the Netherlands. However there are studies that have investigated the effect of certain medicines on stuttering. Pharmacotherapy research will be discussed in this guideline.

2.2.4 Treatment of Children

Therapeutic methods and techniques

Various therapy methods are provided in the Netherlands for the treatment of young children up to the age of six. The Demands and Capacities Model (DCM) is the most commonly applied approach at this moment. DCM treatment is aimed at eliminating or reducing the factors that provoke stuttering or that maintain stuttering. The "demands" placed on the child may focus on the (speech) motor system or may be linguistic, cognitive or social/emotional in nature. Examples include a rapid speech rate in the child's environment, language levels that are too high, a pressured, busy family environment, etc. In addition to this work is undertaken with the child to improve capacities in the same domains ((speech) motor system, linguistic, cognitive or social/emotional).

The Lidcombe Program is another form of therapy offered to young children who stutter. The Lidcombe Program is an operant treatment programme where parents are taught to promote and reward fluent speech in the child and to correct stuttering. This is undertaken in a balanced way using a prescribed approach and a ratio of reward to correction.

There are various other approaches in addition to the methods previously described. Examples of these include the Dell (Dell, 1990) stutter modification programme aimed at children, Riley & Riley's Speech Motor Training (SMT), which improves speech motor planning, as well as social-cognitive behaviour therapy (SCG) (Boey, 2003), which is a programme aimed at reducing the inhibitory emotions and cognitions around stuttering.

In general, parents and the child's immediate environment are closely involved in the treatment. There is always the consideration in the treatment of young children where there is a significant chance of spontaneous recovery. In this situation, no treatment will be provided and the stuttering will be monitored by the therapists or via parental guidance alone in order to prevent the stuttering from developing further.

For older children, depending on the presentation and the outcome of the diagnostic assessment, therapy will focus on enhancing fluent speech and on reducing or eliminating secondary stuttering behaviors, including emotions and cognitions. Just as with adults and adolescents, various therapeutic means and exercise programmes will be deployed for this. These will be adapted to the child's level and needs.

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Chapter 3: What are the Indicators to Treat Children, Adolescents and Adults Who Stutter?

3.1 Introduction

Stuttering is a disorder in the fluency of speech. The first symptoms are usually observed in the first years of life, shortly after language development has started. Stuttering can develop into a communication problem that may severely impair social development and personal well-being. Stuttering may be persistent and difficult to treat at later ages. There is evidence from clinical studies that early interventions enhance the chances of recovery. However some children who start to stutter may recover without any intervention. There is currently insufficient clarity about the number of children who recover spontaneously and it is still not possible to simply predict which children will recover naturally. Recommendations will be formulated in this chapter to enable speech & language therapists and fluency specialists to take evidence-based decisions about at which point to start treating the person who stutters or to allow spontaneous recovery to take place.

3.2 Method

The answer to the principal question of this chapter is based on the limited international publications in the literature about this topic. The support for the recommendation has been primarily based on a single longitudinal study (Yairi & Ambrose, 2005). In methodological terms this study is currently the best one available.

3.3 Evidence

The assessment relates to the question: When does treatment need to take place?

The issue of *spontaneous recovery* in children plays an important role in terms of providing advice regarding this question. Some of the children who have started to stutter recover from this in full. However, figures about spontaneous recovery show large differences varying between 50 and 94% (Yairi & Ambrose, 2013). The "reliability" of these figures is questionable given the different definitions of stuttering used by different authors, the number of years in which the children were followed-up and whether or not the children underwent therapy. The fact that these figures relate to recovery in the entire population should also be taken into consideration. This implies that they are applicable to children who are identified as children

who stutter at the screening phase. The question is whether the recovery percentages are also applicable to the clinical population, in other words, children whose parents have opted for the help of a speech & language therapist or a fluency specialist.

3.3.1 Prevalence and Incidence

The majority of children start stuttering aged between 2 and 4. Although there are case studies of people who started stuttering as teenagers, stuttering occurring after age nine is rare. The risk of developing a stutter appears to be 5% after the age of 4 (Yairi & Ambrose, 2013). Children who started stuttering after age 4 appear to have a greater risk of persistent stuttering (Yairi & Seery, 2011).

The chance of spontaneous recovery decreases in proportion to the amount of time stuttering has been present. Shortly after the first stuttering symptoms have been observed the chance of spontaneous recovery is estimated at approximately 75%. This risk drops to 63% after one year, 47% after two years, 16% after three years and ultimately drops to 5% four years after the stutter started (Yairi & Seery, 2011). The number of children completely recovered one year after starting stuttering is estimated at 6.3 to 9% (Yairi & Ambrose, 2005; Reilly et al., 2013). Recovery of stuttering remains possible in adolescents and adults, but is still relatively rare (Finn et al., 2005; Kell et al., 2009).

3.3.2 Family History

The strongest and earliest predictor of spontaneous recovery is the presence of persistent stuttering or recovered stuttering in the family. If persistent stuttering is present in the family of the child who stutters then the chance of the child continuing to stutter is 65% without treatment. The presence of recovered stuttering in the child's family predicts that the chance of spontaneous recovery is also 65% (Yairi & Ambrose, 2005; Seery & Yairi, 2011).

3.3.3 Gender

From an early stage, there are more boys who start to stutter than girls (2:1). Recovery rates are greater in girls than boys. The ratio of men to women who stutter in adults is approximately 4:1. Girls not only have a more favourable prognosis in terms of recovery, but also appear to recover more quickly than boys. Where stuttering in girls has not been clearly

reduced one year since onset, then the risk of persistent stuttering increases significantly as compared to boys (Yairi & Seery 2011).

3.3.4 Stuttering Symptoms

The severity of stuttering, presence of extensions, blocks and additional physical behaviours during the first year are *not* predictors of persistent stuttering or for recovery of the stuttering (Yairi & Ambrose, 2005; Yairi & Seery, 2011).

A significant reduction in the number and the severity of stuttering symptoms during the first year after stuttering has started, is an important indicator for a potential spontaneous recovery. A reduction in the number of repetitions, the number of units per repetition, blocks and extensions and additional physical behaviours, as well as a reduction in the rate of repetitions are positive predictors of recovery. The reduction is of greater importance than the absolute frequency of the dysfluencies (Yairy & Seery, 2011).

Yairi & Ambrose (2005) observed a reduction in the average weighted Stuttering Like Dysfluencies (SLD) score of more than 50% during the first year to a year after stuttering arose in the group of children whose stuttering recovered after the first stuttering symptoms. The weighted SLD-score has been developed by researchers in order to distinguish mild, but clear stuttering from normal dysfluent speech. Blocks and extensions, which rarely occur or do not occur at all in children who do not stutter, count double in the weighted SLD-score, while repetitions are weighted by the number of units (for instance, m-me counts once, and m-m-m-me counts for 3). In contrast to that observed in those children whose stutter recovered spontaneously, the average weighted SLD-score barely decreased in the group of children who remained stuttering half a year to a year after the stutter arose.

The severity of stuttering, as rated by parents and the researcher, dropped on an 8-point scale of stutter severity (where 0 = normal, fluent speech and 7 = very severe stutter), by an average of approximately 2 points half a year to a year after stuttering onset in the group of children who eventually recovered (Yair & Ambrose, 2005). This decrease virtually corresponds with a reduction of stuttering by one degree of severity on the Yairi & Ambrose (2005) stuttering severity scale. The average severity of stuttering in those children who were still stuttering five years after the start of their stutter, decreased in the first year by an average of less than 1 point. The average scores of parents and the researcher were virtually the same.

Interval	Stuttering severity
0-1	Normal fluent speech
1-3	Mild stuttering
3-5	Moderate stuttering
5-7	Severe stuttering

Stuttering Severity Scale (Yairi & Ambrose, 2005)

An increase in stuttering lasting a period of one to three months occurring one year after stuttering onsetis a signal that the risk of persistent stuttering is increasing (Yairi & Seery, 2011).

The severity of stuttering one year after its onset is also a predictor of persistent stuttering, in contrast to the period of the first 12 months after stuttering has started. The risk of persistent stuttering increases in proportion to severity, rather than absolute frequency (Yairi & Ambrose, 2005; Yairi & Seery, 2011).

3.3.5 Phonological Development

A child's poor phonological skills, that is: the presence of unusual phonological processes and too many common phonological processes for the child's age, may be a risk factor during the initial phase of stuttering. Phonological skills lose their predictive value for the risk of persistent stuttering two years after the onset of stuttering (Yairi & Seery, 2011). Phonological development is not a strong risk factor. The speech & language therapist or fluency specialist should be alert to other risk factors for stuttering where a child's phonological skills are below average (Yairi & Seery, 2011).

3.3.6 Receptive and Expressive Language

Expressive vocabulary at age two is a weak predictor for stuttering arising at age three and is not predictive of stuttering at age four (Reilly et al., 2013). The predictive value of expressive language skills is as yet unclear for persistent stuttering (Yairi & Seery, 2011).

3.3.7 Additional Disorders

Additional disorders may complicate the stuttering problems and, in combination with other risk factors, may increase the chance of persistent stuttering. The predictive value of additional disorders for persistent stuttering is unknown (Yairi & Seery, 2011).

3.3.8 Temperament and Social/Emotional Well-Being

There is insufficient scientific evidence in terms of the effect of an awareness of stuttering or the emotional response of the child to the stuttering as risk factors for persistent stuttering (Yairi & Seery, 2011). Reilly et al. (2013) did not find any convincing evidence that quality of life in the emotional and psychosocial domains differed in a group of three and four year olds who stuttered from children who did not stutter. Furthermore no difference was found in temperament between the two populations (Reilly et al., 2009; Reilly et al., 2013). However, research in the clinical population into the temperament in children who stutter have - on average - a higher level of (negative) reactions and lower self-regulation (Eggers, 2012).

Parental anxiety, anxiety in the child or an agitated environment around the child may at an individual level lead to conscious and/or subconscious verbal and non-verbal reactions to the stuttering. These reactions may contribute to more cognitive and emotional responses in the child which can lead to or reinforce a negative learning process in the child (Korrelboom, 1993). These learning processes are influenced by the child's temperament.

3.3.9 Therapy

Treating young children who stutter in the 15 months after stuttering has started appears to be more effective than starting treatment more than 15 months after stuttering onset. Ingham and Cordes compared the results of different studies investigating the treatment of children who stutter. Of the group of children who had been treated within the 15 months after the stuttering had started, 85.7% achieved the criteria for stutter recovery. The recovery percentage of the group of children receiving therapy later than 15 months after the start of the stutter was 59.4% (Ingham & Cordes, 1998).

A comparable difference was found when the therapy results of children receiving treatment before age six was compared with the results of children whose therapy started after the age of six. Of the young children, 81.8% achieved the criteria for a positive result defined by Ingham and Cordes compared to 54.2% of the older children.

The risk of recurrence in children who received treatment after the age of six appears to be greater than for those children treated before their sixth year (Lincoln et al., 1996; Koushik et al., 2009).

There is evidence that delaying treatment by a year or more does not impact negatively on the treatment duration (Yairi & Seery, 2011). Children who suffered from stuttering longer than 12 months required less time to complete the first phase of the Lidcombe Program than children treated shortly after the beginning of stuttering (Jones et al., 2000; Kingston et al., 2003).

3.4 From Evidence to Recommendations

Quality of evidence

Spontaneous recovery and the *effectiveness of treatment* play an important role in providing advice regarding the question of when a person who stutters needs to be treated. Figures about spontaneous recovery show large differences and risk factors for persistent stuttering have only been investigated to a limited extent.

Treating children before age six has a significant effect on the frequency of stuttering and there is a reasonable certainty that the estimate of this effect size corresponds to the actual effect size. Treatment of older children, adolescents and adults is less effective in reducing the percentage of stuttered syllables. There are insufficient data available to determine how effective treatment of stuttering is on outcome measures such as quality of life, avoidance behaviour, participation and naturalness of speech.

Balance of desired and undesired effects

Stuttering is a disorder in the fluency of speech that can develop into a communication problem which may severely impair social development and personal well-being. Treatment is effective, as well as intensive. Effectiveness in reducing the number of stuttered syllables in the short and medium term is greatest where treatment is applied before age six. Stuttering usually arises before age five. A large proportion of children (50 to 94%) who start to stutter recover from this fully without explicit treatment. A year of carefully monitoring children from the point at which stuttering starts ensures that children with a high risk of persistent stuttering receive treatment at the time that is most effective; this also minimises the treatment of children who may potentially recover spontaneously.

Monitoring

Monitoring is an active process where the speech & language therapist or fluency specialist systematically follows the progress of the stuttering. After the diagnosis / consultation, the speech & language therapist or fluency specialist will train parents in the focused observation and recording of the child's stuttering symptoms. The speech & language therapist or fluency specialist will continue training the parents until they reach an agreement about the severity of the stuttering observed. The use of recording system to be used, the frequency of recording, as well as the mode and frequency of reporting the recorded data back to the speech & language therapist or fluency specialist will be discussed and agreed in consultation with the parents. During the monitoring process the speech & language therapist or fluency specialist will check whether the mode of assessing stuttering severity by the parents continues to agree with his/her own assessment. A review assessment will be undertaken at least once every six months. These review assessments should preferably take place six months and one year after the child has started to stutter.

Values and preferences

a. People who stutter

Given the potential impact of stuttering on the personal and social life of people who stutter and the beneficial therapy results at a young age, the early identification and subsequent referral to a speech & language therapist or fluency specialist is of importance. Parents of children who stutter regularly report that they were referred too late to a speech & language therapist or fluency specialist. The Stuttering Screening List (SSL) is the most commonly used instrument in the Netherlands for the early identification of stuttering. The SSL is freely available via various websites.

Parents and professionals who have contact with young children need to be well informed about the risk signs for persistent stuttering and about the advantages and disadvantages of delaying or starting treatment under the supervision of a speech & language therapist or fluency specialist.

Parents of older children who stutter, adolescents who stutter and adults who stutter searching for treatment should be informed about the options for therapy given the chance of spontaneous recovery of the stuttering may be very limited at these ages. The impact that stuttering has on the personal and social life of the person who stutters may be minimised through treatment.

b. Speech & language therapist - fluency specialists

The early identification of children who are at risk of persistent stuttering is the first step in the process of monitoring and potential treatment of stuttering. Speech & language therapists and fluency specialists are the designated experts to inform healthcare providers, teachers and supervisors of nurseries and child daycare facilities about the risk signs of persistent stuttering and to provide them with the necessary means. It is important that the speech & language therapist / fluency specialist is expertly skilled in the monitoring of young children who stutter.

The multifactorial character of stuttering, the potential impact of stuttering on the life of the person who stutters and the limited effect of treatment on recovery from stuttering should be taken into consideration in determining whether treatment of an older child, adolescent or adult is indicated. The speech & language therapist or fluency specialist should detail each of the ICF elements in this process.

c. Costs

There is insufficient information available about the costs of therapy. There are, however, data available about the average number of treatment sessions in the Lidcombe Program. The average number of treatment sessions to achieve phase II of the Lidcombe Program (that is: a significantly reduced stutter severity) is 10 sessions for mild stuttering, 12 sessions for moderate stuttering and 14 sessions for severe stuttering (Koushik et al., 2011). The median number of treatment sessions during phase 1 is 15.4, after which another 10 treatment sessions will be provided during the stabilisation phase (Reilly et al., 2013).

Recommendations

The recommendations below should be considered in their entirety.

1. The treatment of children who begin to stutter before the age of four, starts before they become five years of age.
2. The speech & language therapist or fluency specialist will monitor the child who started to stutter, before four years of age, for signs of spontaneous recovery during a period of one year after onset.
3. The speech & language therapist or fluency specialist will start treatment 12 months after onset, when the severity of stuttering has not clearly* decreased in the last 3-6 months.
4. The speech & language therapist or fluency specialist will take into account the recovery pattern of stuttering in the family, gender, phonological skills, additional disorders and the temperament of the child in the decision to start intervention or to monitor fluency development.
5. The speech & language therapist or fluency specialist will start intervention (even if the child has been stuttering for less than 12 months) if the child suffers from

stuttering, if the parents are concerned about the child's stuttering or if the child is showing reluctance to communicate.

6. Parents of a child under 6 years of age fill in the Stuttering Screening List (SSL) to determine whether further diagnostic assessment by the speech & language therapist or fluency specialist is indicated.

7. The speech & language therapist or fluency specialist will inform the medical professionals, teachers and supervisors at day care centers within his/her network about the purpose and usage of the SSL and he/she will provide information about relevant websites.

8. In cases of children over 6 years of age, adolescents or adults, seeking help in stuttering, the speech & language therapist or fluency specialist will, together with the client, make an inventory of ICF-elements (International Classification of Functioning, Disabilities and Health) to find out whether treatment is indicated.

* a clear decrease in stuttering corresponds to an average reduction of at least 2 points on the 8-point stutter severity scale (Yairi & Ambrose, 2005) or an average reduction of 2 points on the Lidcombe Program stuttering severity scale. This decrease in severity needs to be observed by both the parents and the speech & therapy therapist / fluency specialist.

Rationale for the recommendations

- Stuttering may have severe consequences on the quality of life of children and adults who stutter.
- Treating young children who stutter is more effective than treating children who are six or older: the risk of a recurrence is smaller and the chance of persistent recovery is greater (Ingham & Cordes, 1998; Lincoln et al., 1996; Koushik et al., 2009).
- Treating stuttering within the 15 months after it has started has a higher chance of recovery than starting treatment once the stuttering has been present for more than 15 months (Ingham & Cordes 1998).
- Observing the first symptoms of stuttering after the age of 4 or 5 has been associated with an increased risk of persistent stuttering (Yairi & Ambrose 2005; Seery & Yairi 2011).
- A large group of children who start to stutter will recover fully without treatment. However, there are large differences in the figures for spontaneous recovery (Yairi & Ambrose 2013; Onslow & O'Brian 2013).
- Delaying treatment for a year does not extend the length of treatment required (Jones et al., 2000; Klingston et al., 2003).
- The risk of persistent stuttering increases if stuttering is still present one year after the first symptoms have been observed (Yairi & Ambrose 2005; Seery & Yairi 2011).
- A downward trend in stutter severity during the first year after the first stuttering symptoms have been observed is a clear sign of a potential recovery.
- A reduction in the number of repetitions, the number of units per repetition, blocks and extensions and additional physical behaviours, as well as a reduction in the rate of repetitions are positive predictors of recovery (Yairi & Ambrose 2005; Seery & Yairi 2011).

- The severity of stuttering in the first year is not a predictor of persistent stuttering or predictive of a recovery in stuttering (Yairi & Ambrose, 2005; Seery & Yairi, 2011).
- Stutter severity, the presence of secondary behaviour, blocks and extensions are predictors of persistent stuttering if the stutter has been present for a year or more (Yairi & Ambrose 2005; Seery & Yairi 2011).
- Therapeutic intervention is indicated if the parents / carers are concerned (Reilly et al., 2013).
- The average weighted SLD score decreases by more than one half in the group of children who recover from stuttering in the six months to a year after the beginning of stuttering. The average weighted SLD score decreases minimally in the group of children who continue to stutter in the six months to a year after stuttering has started. A comparable trend is evident for stutter severity (Yairi & Ambrose, 2005).
- If persistent stuttering is present in the family of the child who stutters then the chance of the child continuing to stutter is 65%. The presence of recovered stuttering in the child's family predicts that the chance of spontaneous recovery is also 65% (Yairi & Ambrose, 2005; Seery & Yairi, 2011). The ratio of boys to girls who stutter at a young age is 2:1. The ratio of men to women who stutter in adults is approximately 4:1. Girls recover more quickly. If there is no clear recovery observable in a girl who has been stuttering for more than one year then the chance of persistent stuttering increases (Yairi & Ambrose 2005; Seery & Yairi 2011).
- Additional disorders, a higher negative reaction and lower self-regulation may complicate the stuttering problems and increase the chance of persistent stuttering.
- Parents of children who stutter regularly report that they were referred too late to a speech & language therapist or fluency specialist.
- The SSL is the most commonly used screening instrument in the Netherlands. The SSL is freely available via various websites.

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Chapter 4: What is the Diagnostic Value of Tests for Children and Adults who Stutter?

4.1 Introduction

Stuttering is considered to be a multifactorial problem. Diagnosing stuttering is more than just establishing whether or not stuttering is present. During the assessment phase, the speech & language therapist needs to explore the various ICF domains (functions, external factors, contextual factors, personal factors, participation and activities) in more depth and detail, then analyse and interpret their mutual associations.. In order to realise as complete a picture as possible, the speech & language therapist will use the best available diagnostic instruments that are applicable and will draw in all the individuals from the client's environment who are relevant to the diagnostic process. The value of the instruments most commonly used in the Netherlands is discussed in this chapter.

There are many examples in the medical domain where a so-called golden standard - often a histological test - is available to which the outcomes of a screening or diagnostic test can be compared (Biddle, 2002; Rutjes, 2007). This golden standard is the (valid) instrument by which to ascertain a diagnosis with certainty. Discussions about the diagnostic test can be limited to its reliability and the criterion validity (see below). The concept of test accuracy is often used instead of criterion validity. This accuracy may be expressed as positive and negative predictive values in regards to sensitivity or specificity. A negative result for a highly sensitive test means the condition can be excluded. A positive result to a highly specific test means the condition is present.

However, in the absence of a golden (or silver or potentially copper) standard, there is no longer a "true" value and the diagnostic accuracy paradigm no longer applies. In cases such as these the paradigm of various aspects of validity becomes relevant. The important concepts here are the content validity, criterion-related validity and construct validity of a test or instrument.

Content validity

This relates to the degree in which the items of a test adequately represent the dimension(s) to be measured. This evaluation is often subjective, in other words it is carried out by experts who then assess whether the items are appropriate.

Criterion-related validity

This relates to how effective the instrument is in predicting someone's performance during specific activities, such as their performance on a different related instrument (concurrent validity) or future behaviour (predictive validity).

Construct validity

This relates to the degree in which a test measures a theoretical concept or characteristic. A distinction may be made between convergent and divergent validity.

In terms of convergent validity test developers are interested in the question as to whether an instrument correlates strongly with variables or the characteristics it should be correlating with. For instance, an instrument measuring articulation should be correlated strongly with another instrument measuring articulation.

Divergent validity is present when an instrument *does not* correlate with variables which it should differ from.

An instrument should also be reliable in addition to being valid. The following concepts are of importance here: internal consistency, test-retest reliability, intra-rater reliability and inter-rater reliability.

Internal consistency

Internal consistency (inter-item consistency) measures how well individual items (or questions) in a scale or scales agree with the composite scores.

Test-retest reliability

Test-retest reliability measures the consistency in scores at two different points in time.

Intra-rater reliability

Intra-rater reliability measures whether the researcher or the test instrument allocated the same scores to an individual in situations where the test is performed on multiple occasions.

Inter-rater reliability

Inter-rater reliability measures whether two observers or researchers score an individual in the same way.

In this chapter the validity and reliability of a number of instruments that are in use or of interest in the Netherlands will be discussed. The following instruments will be covered:

- **Stuttering Screening List** and **Stuttering Detection Instrument**, both used to flag / detect stuttering
- **Communication Attitude Test -DR**,
- **Test for Stuttering Severity (TfS) readers** / **non-readers** and **Stuttering Severity Instrument** (SSI- 3/4), both used for determining the severity of *stuttering*
- **Behaviour Assessment Battery (BAB)** including **Communication Attitude Test** and **Erickson-S24** and **Overall Assessment of Speaker's Experience of Stuttering (OASES),** both used to detail various aspects of the *quality of life*.

TfS, SSI-4, OASES and BAB also measure various aspects of *avoidance behaviour*.

4.2 Method

A search for relevant studies was undertaken both in bibliographical databases (Medline, Cinahl, Cochrane, Picarta) as well as via Google (see Appendix 5 for the sources used and keywords).

GRADE is not suitable for assessing the quality of diagnostic instruments for which there is no golden standard; this also applies for previous grading systems. Statistical measures are used instead; for instance, values are interpreted in terms of limited, moderate, good or perfect reliability.

Biddle, Watson and Hooper (2002) published criteria in their report "Criteria for Determining Disability in Speech-Language Disorders. Evidence Reports/Technology Assessments, No. 52" for determining the *degree* of validity and reliability of tests or instruments.

They used the following criteria for the various aspects of *reliability*:

- Internal consistency:
    - both Cronbach's alpha coefficient, and the Kuder-Richardson statistic (K-R 20), should be at least 0.80 for there to be good internal consistency;

- Test-retest and intra-rater reliability:
  - at least 0.80 in the case of a correlation coefficient, or at least 0.70 in the case of Cohen's kappa;
- Inter-rater reliability:
  - at least 0.90 in the case of a correlation coefficient, or at least 0.70 in the case of Cohen's kappa;

The following criteria were used by Biddle, Watson and Hooper (2002) for the various aspects of *validity*; *all* criteria need to be satisfied for there to be a valid instrument:

- test developers investigate the relationships between sub-tests, composite scores and total scores, formulating a priori hypotheses about these relationships and scoring patterns;
- these relationships all need to be statistically significant ($p < 0.05$) and
- in the case of correlation coefficients they need to be at least 0.30 (corresponding to a moderate correlation).

In terms of *standardisation* the following criteria were used by Biddle, Watson and Hooper (2002); *all* criteria need to be satisfied for there to be good standardisation:

- data are available about the target population for the instrument in question;
- an adequate sample size needs to have been used (at least 100 per group); and
- evidence has to be provided about how representative the sample is of the population.

4.3 Evidence

a. **Tests for determining stuttering severity**
- **Stuttering Severity Instrument**

The SSI 3/4 is an instrument to determine the severity of observable stuttering behaviours. The instrument includes the following dimensions:

- frequency: expressed as a percentage of stuttered letter groups and converted to a scale with scores varying from 2 to 18,
- duration: the average length of the longest three stutter moments, rounded up to a tenth of a second and converted into a scale with scores varying from 2 to 18,
- physical concomitants: distracting sounds (score 0-5), facial grimaces (score 0-5), head movements (0-5) and movements of the extremities score 0-5), expressed as a total score varying from 0 to 20

There is an absence of information from Riley about the content validity of the SSI-3 (Riley, 1994). There is only indirect evidence available regarding the criterion validity. Neither the reliability nor the standardisation appear to be adequate (Biddle, 2002).

The SSI-4 differs from the SSI-3 in various aspects: *"The SSI-4 attempts to offer more avenues of evaluation, including self-reports and additional beyond clinic and telephone samples."* There is the option of computer calculations of the score, and a score for the naturalness of speech has been added. The validity of this instrument is adequate. The reliability is difficult to assess as percentage agreement has been reported and not kappa statistics. The percentage "accidental" agreement was not taken into account in calculating these percentages. The standardisation was inadequate due to the sample size being too small and not necessarily representative.

The SSI-3 has been translated into Dutch, but not validated. The psychometric properties are not known. The SSI-4 has not been translated into or validated for Dutch practice.

Conclusion

The validity and reliability of the English version of the SSI-4 has been investigated in more detail than the English version of the SSI-3. Its validity satisfies the criteria that may be applied to a properly valid test. This cannot be said of the standardisation, whilst the reliability is difficult to assess due to the absence of kappa coefficients.

- **TfS-R and TfS-NR**

Boey (2000) has published data on the validity and reliability of two tests, namely the Test for Stuttering Severity in readers (TfS-R) and Test for Stuttering Severity in non-readers. There is no description of how the children were recruited and selected. Data were obtained for the TfS-R from 177 clients aged 8 years and 1 month up to 53 years and 9 months, which were used as the basis for the standardisation. The gender ratio was not reported for these 177 clients. Data were obtained for the TfS-NR from 342 children aged from 2 to 7, which were used as the basis for the standardisation. The gender ratio was 256:86 according to Boey[3].

There is very little methodological information for both tests about the content validity, whilst concept validity is not discussed. According to Biddle's criteria (Biddle, 2002), hereafter referred to as the BWH criteria, the validity of these tests has not been *proven.*

The reliability of these in terms of the BWH criteria also leaves a lot to be desired. Information about the internal consistency is absent, whilst the test-retest reliability does not exceed the threshold for reliability. In respect to the TfS-NR the inter-rater reliability is not at the required level either. In terms of the standardisation of both tests it may be noted that the representativeness of the sample cannot be assessed.

Conclusion

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[3] which must be an error as 256 + 86 = 352 and not 342.

There is only a moderate amount of information about the content validity. The criterion validity of the TfS-R / TfS-NR appears sufficient. There is no information available about concept validity. Reliability and the standardisation do not appear to be adequate in all aspects.

b. Tests for identifying stuttering

The two tests to be described and used for identifying stuttering - the Stuttering Screening List and the Flemish Instrument for Detecting Stuttering have both been derived from a test instrument developed by Riley & Riley, and therefore display a strong agreement. They will be discussed separately due to their existence as individual instruments.

- **Stuttering Screening List**

The Stuttering Screening List (SSL) is an instrument which is aimed at determining whether or not there is (the onset of) stuttering or whether the child has a risk of persistent stuttering and whether they need immediate help. Parents are asked to complete a form with six questions. The Association of Stuttering Centres Netherlands has described this instrument as "very reliable and valid", and refers to the Institute for New Stutter Research (INSO) in Zutphen for more specific information. However this information is not accessible, nor available.

The Stuttering Screening List (SSL) is based on an American test instrument (Riley, 1989). The items described in this screening list came about as follows. Items from the Stuttering Prediction Instrument (SPI) were tested with 8 children (4-7 years) who had been accepted for stuttering therapy and 31 children who were being monitored but did not receive therapy. None of these 31 children appeared to require therapy within two years. In order to calculate the percentage of false positives, Riley and Riley subsequently compared the scores between the children in therapy and the children not receiving therapy. These percentages varied from 0% ("phonatory arrests") to 12.6% ("onset more than 12 months ago"). That is to say that none of the children with "phonatory" arrests were erroneously in therapy, but 126 in 1,000 children with faltering speech for over 1 year would have been unnecessarily referred for therapy. The tested items were defined in layman's terms and provided within categories in a screening list with the advice to refer to a fluency specialist if three or more symptoms are present.

- **Stuttering Detection Instrument**

According to Stes and Boey (1997), the Stuttering Detection Instrument (SDI) is: "an instrument that enables the selection of young children who are potentially stuttering and also provides indications about the severity of stuttering and the risks of chronic stuttering. It is, in essence, a simple, brief questionnaire completed by parents, potentially with the help of a healthcare professional".

The sample used to investigate the SDI was consisted of children (N=42; average age: 68 months) who stuttered and who received treatment at the Centre for Stuttering Therapy in Antwerp. The gender ratio in this group was 34:8. For children who did not stutter (N=42; average age: 67 months), the recruitment and selection process has not been described. The gender ratio in this group was the same as the researchers consciously aimed for this.

In terms of validity Stes and Boey (1997) only provide information about the criterion validity using the Stuttering Prediction Instrument (SPI) as the "golden standard". How the assessment to determine the criterion validity was carried out is not described in detail: for instance, was the researcher who investigated whether there was stuttering present using the SPI aware of the outcome of the Stuttering Screening List (SSL) (also referred to as the Stuttering Detection Instrument) and *vice versa?* Irrespective of this point, Stes and Boey ascertained a high degree of sensitivity and specificity for their SDI. However, the positive predictive value of the test did not appear to be as high: 0.63 and 0.55 at a threshold of SPI≥10 and SPI>13, respectively. The corresponding negative predictive values were 0.98 and 1.00 respectively. The test did appear to perform better at excluding rather than indicating stuttering, whilst more than 10% of the children were incorrectly referred for stuttering. Stes and Boey do not provide any information about the various aspects of reliability and standardisation.

Conclusion

There is insufficient information available or known about the validity and reliability of the Stuttering Detection Instrument and Stuttering Screening List in order to arrive at a judgment regarding either the validity or reliability of this instrument.

c. Tests for determining speech attitude

In 1985 Brutten, developed the Communication Attitude Test (CAT). This original American CAT was subsequently translated into Dutch as the Communication Attitude Test - Dutch (CAT-D). According to Bremer et al. (1993) it appeared that "the language use of the items imposed significant demands on the children's language level". These authors developed a simplified version under the name Communication Attitude Test-Dutch Revised (CAT-DR). The rationale of this instrument is according to Bremer et al.: "increasingly more authors are demonstrating that communication attitudes in children do impact on their fluency problem". The CAT-D comprises 32 items with statements or expressions regarding speech. The CAT-DR contains sentences with negations and amendments that have been introduced in word choices and sentence constructions. The CAT-D and CAT-DR were compared in a group of 162 children from the Dutch city Almere with *normal speech* aged between 6 and 13. The comparison reviewed the test-retest reliability, concept validity, internal consistency and standardisation. In terms of the CAT-DR the test-retest reliability was considerably better than the CAT-D. The internal consistency of the CAT-DR was good, and somewhat better than the CAT-D. In terms of concept validity there was good agreement between the CAT-D and CAT-DR. Other aspects

of validity and reliability were not investigated. In respect to the standardisation there was no agreement between the CAT-D and CAT-DR, given that for the CAT-DR the scores differed by age as well as gender. The number of children in the study categorised by gender and age was insufficient for it to be classified as an adequate standardisation. This was further compounded by the fact that all the children were recruited from Almere alone.

Conclusion

The construct validity of the CAT-DR appears to be adequate. No information is available about the other aspects of validity. Reliability appears to be adequate. However, this does not apply for the standardisation.

d. Tests for obtaining a multi-dimensional view of speech problems and speech-related problems

- **Behaviour Assessment Battery (BAB) for adults**

The BAB – a multi-dimensional research instrument developed by Brutten and Vanryckeghem (2003) that investigates the behavioural, cognitive and affective components of stuttering - comprises three self-reporting based test procedures: the Speech Situation Checklist (SSC), the Behaviour Checklist (BCL) and the Erickson S-24 (S-24). The SSC evaluates the degree to which a client experiences a speech situation as anxiety-inducing and disruptive to speech. The SSC consists of two components: the SSC-ER and the SSC-SD. The first component attempts to map out the degree of emotional reactions someone experiences in 55 speech situations. The second component is aimed at measuring the degree of speech disruption related to these speech situations. The BCL was developed to measure the number and frequency of behaviours that a person who stutters consciously applies as a way of dealing with (the anticipation of) the occurrence of dysfluencies. This therefore relates to avoidance behaviour. The S-24 / CAT measures the client's attitude in respect to speech and communication.

The BAB has a number of evident benefits according to Dines (2011):

- there have been very positive reviews about the BAB;
- the BAB has been used in numerous and diverse cultural settings in order to validate the tests;
- the information obtained with the BAB enables the healthcare practitioner to target therapy according to the client's needs.

Dines (2011) has also noted the high costs and time required to take all the tests as disadvantages.

The composition of sample used to investigate the SSC was as follows: There were 41 people who stuttered who were recruited and 155 persons who did not stutter. The ages of the first group varied from 17 to 50, and those of the second group from 18 to 50. The gender ratio was 32:9 for the first group and 73:82 in the second group. These ratios correspond well with those found in the population.

Brutten and Vanryckeghem investigated various aspects of the validity and standardisation of the *SSC-ER and SSC-SD*. In terms of the reliability these researchers restricted themselves to assessing the internal consistency. It is not clear, however, why the test-retest reliability was not evaluated.

Conclusion

The validity of the SSC is adequate as demonstrated by the positive findings in respect to concept validity, content validity and criterion validity. Reliability has been partly investigated. The standardisation appears adequate.

Brutten and Vanryckeghem *did not investigate the reliability of* the BCL. The arguments for not doing so only relate to investigating the internal consistency. It is not clear, for example, why the test-retest reliability was not evaluated. All facets of validity have been extensively investigated.

Conclusion

The validity of the BCL is adequate proven by the positive findings in respect to concept validity, content validity and criterion validity. Reliability has not been investigated.

Brutten and Vanryckeghem *only investigated the reliability of the S-24* in terms of the internal consistency component. This was adequate. All facets of validity were extensively investigated and deemed to be good. In terms of standardisation it may be noted that gender does not play a role in the S-24 scores. This applies both for people who stutter and those who do not stutter.

Conclusion

The validity of the S-24 is adequate as evidenced by the positive findings in respect to concept validity, content validity and criterion validity. Reliability has been partly investigated. The standardisation appears adequate.

- **Behaviour Assessment Battery (BAB) for children**

The BAB for children consists of the same three components as the adult version except that the Communication Attitude Test (CAT) replaces the S-24. The benefits and disadvantages of the BAB, as described by Dines et al. (2011) have been previously discussed.

The sample used to investigate the BAB for children was composed as follows: 270 children were recruited from the St Michiels School in Merelbeke, Flanders. The gender ratio was 145:126. The age of these children varied from 7 to 12 years, with 9 years and 3 months as the average. According to the researchers this was a representative group of children for Flanders. Ninety children were recruited from clinical centres and private practices distributed across Flanders. The average age of these children was 9 years and 4 months. The gender ratio was 70:20.

In terms of the reliability of the *SSC* the researchers restricted themselves to assessing the internal consistency. As to why the test-retest reliability was not investigated is not explained by the researchers. The internal consistency of the SSC was good. That also applied to the various aspects of validity. An important finding in the study was that the gender of the children who stuttered did not have an effect on the scores on the SSC-ER and the SSC-SD, however this did not apply for children who did not stutter: girls scored higher on the SSC-ER and SSC-SD compared to boys.

Conclusion

> The validity of the SSC-ER and SSC-SD in children is adequate as shown by the positive findings in respect to concept validity, content validity and criterion validity. Reliability has been partly investigated. The standardisation appears adequate.

Brutten and Vanryckeghem *did not investigate the reliability of* the BCL. The arguments for not doing so only relates to investigating the internal consistency. The test-retest reliability was not evaluated and it is not evident as to the reason why. All facets of validity were extensively investigated and may be described as good. In contrast to the SSC, no gender effect was found for the BCL scores.

Conclusion

> The validity of the BCL in children is adequate as evidenced by the positive findings in respect to construct validity, content validity and criterion validity. Reliability has not been investigated. The standardisation appears adequate.

Brutten and Vanryckeghem *investigated various aspects of the reliability of the* Communication Attitude Test (CAT), such as internal consistency and test-retest reliability.

Both of these aspects point towards a reliable test. The researchers also discussed sensitivity or the discriminatory capacity of the test in the context of test-retest reliability. They report a Swedish study which appeared to demonstrate that scores after therapy were significantly lower compared to the children who were on the waiting list for therapy. All facets of validity were extensively investigated and may be described as good. The school children's CAT scores were not influenced by gender.

Conclusion

The validity of the CAT in children is adequate as evidenced by the positive findings in respect to construct validity, content validity and criterion validity. Both reliability and standardisation appear to be adequate.

- **Overall Assessment of the Dutch Speaker's Experience of Stuttering for adults (OASES-A-D)**

The work carried out by Koedoot (2011) in the Netherlands is important in terms of the psychometric evaluation of the Dutch translation of the Overall Assessment of the Speaker's Experience of Stuttering for adults (OASES-A-D). OASES-A-D is a self-reporting based instrument consisting of four components:

1. general perspectives about stuttering (20 items)
2. affective, behavioural and cognitive reactions to stuttering (30 items)
3. functional communication difficulties (25 items)
4. impact of stuttering on the speakers quality of life (25 items)

Koedoot (2011) involved 142 individuals in the research. The data were partly obtained from another study, and partly through contributions from speech & language therapists / fluency specialists who had asked their clients to complete the OASES-A-D. University educated individuals were overrepresented in this sample: 50% compared to 27% in the general population, which undermines the generalisability of the research results. The gender ratio was 101:37.[4] The average age was 34.5 years and age varied from 18 to 74.

In terms of reliability Koedoot (2011) only evaluated internal consistency; which was categorised as good. Test-retest reliability was not evaluated. In terms of validity, criterion and concept validated were assessed. Content validity was therefore not assessed. The results in terms of the criterion validity were good given the strong correlations between the impact scores of the OASES-A-D and the various other instruments, such as the Dutch version of the S-24. In terms of the concept validity of the OASES-A-D there were a number of shortcomings. For instance, Koedoot et al. (2011) note: ... all sections of the OASES-A-D questionnaire were able to discriminate between groups of participants with different stuttering severity levels

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[4] Four people were excluded because they were younger than 18.

(..) with the exception of discriminating between participants with moderate and severe stuttering as assessed by the SA scale *(SA is a self-assessment scale).*

Conclusion

> The reliability of the OASES-A-D has not as yet been sufficiently investigated to designate this instrument as a valid and reliable diagnostic instrument. Information about standardisation is absent.

- **Overall Assessment of the Dutch Speaker's Experience of Stuttering for children at primary school and adolescents (OASES-S-D and OASES-T-D)**

Lankman, Yaruss & Franken (2015) validated and evaluated the Dutch translation of the OASES-S and OASES-T by way of a scientific placement in the context of medical training. Both instruments have the same four components as the OASES-A, as described above. The OASES-S consists in total of 60 questions and the OASES-T has 80 questions. Lankman recruited 145 trial participants for her study via speech and language therapists and fluency specialists across the Netherlands, and used school pupils who did not stutter (N=82) as a control group. Children from a primary school in Leiden were approached for the control group. Although the recruitment method is extensively described it is unclear where the teenagers aged 13 to 17 were recruited from.

The OASES-S was completed by 152 children and the OASES-T by 75. The gender ratio for the group who stuttered and who completed the OASES-S was 71:30; for those who did not stutter the ratio was 23:28. The corresponding ratios for the group of teenagers who completed the OASES-T were 35:8 and 18:13 respectively.

In terms of the reliability Lankman only investigated the internal consistency of the OASES-S and OASES-T. Test-retest reliability was not investigated. In terms of validity, Lankman investigated the criterion and concept validity. Content validity was therefore not assessed.

The internal consistency of the OASES-S is inadequate. This is in contrast to what Lankman reports. She set the threshold for Cronbach's alpha at a value of at least 0.7 in order to have good internal consistency. According to the BWH criteria this should be at least 0.80; these criteria are specified here.[5] On the basis of the findings for the criterion and concept validity the validity may be designated as good. There were some shortcomings in terms of the

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[5] According to Biddle AK, Watson LR, Hooper CR, et al. (2002) the criterion for reliability is "strictly" met if the condition is met: Internal consistency reliability, measured using either Cronbach's coefficient alpha or Kuder-Richardson statistics (K-R 20), is greater than or equal to 0.90. However they add: "Some might reasonably argue that the criterion for internal consistency reliability is set too high given the complexity of speech and language functioning and their corresponding disorders. Additionally the resultant variability in daily performance suggests that our criterion for test-retest reliability or intra-rater reliability also may be too high. Thus, we defined a "relaxed" criterion, which differs from the strict criterion in that internal consistency reliability may be as low as 0.80 and/or test-retest/intra-rater reliability may be as low as 0.80 (correlations) or 0.70 (Cohen's Kappa)."

standardisation. The internal consistency of the OASES-T is adequate. However details regarding standardisation are absent. Both the concept and criterion validity are sufficient according to the positive findings provided by Lankman.

Conclusion

> The reliability of the OASES-S-D and the OASES-T-D have not as yet been assessed in detail. The criterion and concept validity are adequate. Information about standardisation is incomplete.

e. General conclusion

No single instrument has had every aspect of validity and reliability investigated. This means that there is no *proven* valid and reliable test available with adequate standardisation.

In terms of validity the (Dutch, Flemish version of the) Behaviour Assessment Battery for children and adults has been studied the most and has good scores on all aspects of validity. In terms of internal consistency - one aspect of reliability - the various components of the Behaviour Assessment Battery (children and adults) are also reliable. Other aspects of reliability, such as the test-retest reliability have not been investigated for all of the various components. This is striking given that reliability is one of the first requirements of a test. After all, an instrument that is valid but not reliable, is of no use.

In respect to reliability the test for stuttering severity (readers and non-readers) has been studied the most, but the test is not reliable in every aspect. The validity of this test has only been partly investigated, and was deemed sufficient in terms of criterion validity.

The results of the research into the (Dutch) versions of the OASES for adults, children aged 7-12 and teenagers aged 13-17 revealed that there was good validity, but further research is required for the reliability and standardisation. The various Dutch, language-specific versions of the OASES are not as yet suitable for use in practice.

4.4 From Evidence to Recommendations

Quality of evidence

No single instrument has had every aspect of validity and reliability investigated. This means that there is no fully proven, valid and reliable test available with adequate standardisation. At this point the BAB (children and adults) and the SSI appear to be the most valid and reliable instruments.

Values and preferences

d. People who stutter

Benefits of the SSI 3/4:

- places little burden on PWS with a minimal time investment;
- it can be used both within and beyond practice settings.

Disadvantages of the SSI 3/4:

- The SSI 3/4 is an English-language test, there is no Dutch translation available.

Disadvantages of the BAB (adults and children):

- it is more time intensive;
- it contains Flemish expressions;
- it requires a good command of Dutch.

e. Speech & language therapist / fluency specialists

Benefits of the SSI 3/4:

- it is used internationally;
- it can be used both within and beyond practice settings.

Disadvantages of the SSI 3/4:

- The SSI 3/4 is an English-language test, there is no Dutch translation available.

Benefits of the BAB:

- it has been investigated the most thoroughly and scores well on various aspects of validity and reliability.
- it fits in well with the ICF framework.

Disadvantages of the BAB (adults and children):

- it is more time intensive;
- it contains Flemish expressions;
- it requires a good command of Dutch.

The OASES provides options for the future, but is not as yet available in the Netherlands.

f. Costs

A disadvantage of the Behaviour Assessment Battery (BAB) for children and adults is the high purchase price.

Recommendations

The starting point for the recommendations below is that the speech & language therapist works in accordance with the professional standards for clinical decision making and outcome measurement. It is assumed that the symptoms and presentation have been explored and that a case history interview has been undertaken with the PWS and/or parents/carers.

> 9. The speech & language therapist or fluency specialist determines stuttering severity with a suitable instrument. The Stuttering Severity Instrument (SSI) is preferred because the SSI is used internationally, is valid and seems to be reliable.
> 10. The speech & language therapist or fluency specialist assesses functions, external factors, contextual factors, individual factors, participation and activities according to the ICF principles during diagnostic assessment. The Behavior Assessment Battery is recommended as a diagnostic instrument for children over 6 years, adolescents and adults. For children under 6 years of age no specific recommendation is given due to the lack of specific diagnostic instruments for this age group.

Rationale for the recommendations

- A relatively large weight has been given to the validity, reliability, availability and the link with international developments, and less to factors such as costs, time investment, and the language of the BAB and the English version of the SSI.
- Insufficient information is available about the validity and reliability of the Stuttering Detection Instrument (SDI) and the Stuttering Screening List (SSL). The SSL is the most commonly used screening instrument for stuttering in the Netherlands and is freely available via various websites.

Research recommendations

The test-retest reliability and standardisation of the various age-specific versions of the Dutch OASES need further research.

The test-retest reliability of the majority of components of the Dutch and Flemish versions of the Behaviour Assessment Battery require further research.

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Chapter 5: What are the Effects of Stuttering Therapy in Children who Stutter up to Age Six?

5.1 Introduction

The effectiveness of the Demands and Capacities Model (DCM) and the Lidcombe Program (LP) will be described in this document. In the Netherlands the DCM and the LP are the most commonly employed interventions for treatment of stuttering in children up to age 6. The starting point is that the effectiveness of a stuttering therapy has to be evaluated in a randomised controlled trial (RCT) in intervention studies with a control group. Both studies where a stuttering therapy is compared to "doing nothing", as well as studies comparing various stuttering therapies will be considered.

This document contains the conclusion and summary of the master's thesis: *Evidence-based guideline on stuttering in children and adults. The quality of evidence for the effect of treatment of young children who stutter* (Pertijs, 2013). This relates to a feasibility study where both treatments were compared after 3 months. The effectiveness of Speech Motor Training (Riley & Riley, 1999) and Social-Cognitive behavioural therapy (Boey, 2003) will also be discussed to supplement the conclusion and recommendation in the thesis and in order to sketch as thorough an image as possible of the most commonly used therapies and approaches in the Netherlands.

The draft recommendation for the treatment of young children who stutter up to age 6 will be discussed in the last paragraph of this document.

5.2 Method

Three systematic reviews were selected and assessed *in order to answer the research question as to what the desired and undesired effects are of the Demands and Capacities Model (DCM) compared to the Lidcombe Program (LP) in terms of stuttering severity, avoiding speaking situations, naturalness of speech and quality of life of children who stutter up to age 6* . A meta-analysis (not published in a peer-reviewed journal) was also discussed.

5.3 Evidence

One and the same study (Franken et al., 2005) was discussed in all three systematic reviews included to answer the research question; this study compared the effect of the Demand and Capacities Model directly against the Lidcombe Program. In this study the average percentage of stuttering decreased in the Demand and Capacities Model group by 60.7 % from 7.9% (S.D.= 7.1) to 3.1% (S.D. = 2.1) and the average percentage of stuttering dropped in the LP group by 48.6% from 7.2% (S.D. = 2.0) to 3.7% (S.D. = 2.1). In the systematic review by Nye et al. (2012) the effect calculated in this comparative study amounted to g = -0.275 (95% CI: -1.066 to 0.517). Assuming a threshold value of (-) 0.5 (the threshold value for clinical relevance used by the GRADE working group as standard) the point estimate of -0.275 does not indicate a clinically relevant difference in the reduction of the percentage of stuttered syllables after treatment with the Lidcombe Program compared to treatment based on the Demands and Capacities Model. The lower limit (-1.066) and upper limit (0.517) of the 95% confidence interval both exceed the threshold value of (-) 0.5 for clinical relevance. This implies the possibility that the LP is more effective than the DCM, or conversely that the DCM is more effective than the DCM.

Indirect comparisons of treatment based on the Demands and Capacities Model with the Lidcombe Program by contrasting RCTs where the Demands and Capacities Model and non-treated control was compared to the Lidcombe Program and non-treated control, are not possible due to the absence of RCTs where treatment with the DCM was compared against a non-treated control group.

The systematic review and meta-analysis by Nye et al. (2012) discusses three RCTs where treatment with the Lidcombe Program was compared to a non-treated control group. Assessment of the systematic review and meta-analysis leads to the conclusion that treatment of children who stutter up to age 6 with the Lidcombe Program is more effective in the short and medium term (3 to 9 months) in reducing the percentage of stuttered syllables than when stuttering is not treated. The effect found may be considered to be large (g = 0.97, 95% CI: 0.58 – 1.30, *p* < .001). The evidence for this effect is of moderate quality. This implies that there is some uncertainty about the effect size. The fact that treatment with LP is more effective than no treatment is supported by a meta-analysis published by Onslow et al. (2012). In this meta-analysis the average percentage of stuttered syllables in the Lidcombe Program group was 2.9% lower than the percentage in the control group (95% CI = 1.9 to 3.8, *p* < 0.001). The chance of a stuttering percentage being lower than or equivalent to 1, after 3 to 9 months follow-up, was seven times higher for the group who were treated than those who were not (OR = 7.5, 95% CI = 2.7 to 20.9, *p* < 0.001) [6].

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[6] Stutter severity less than 1 stutter percentage is perceived as normal fluent speech by an untrained listener (Lincoln et al., 1997)

The undesirable effects of treatment with the Lidcombe Program, such as anxiety, aggression, avoidance and depression, and the effect on the quality of the parent-child relationship were investigated by Woods et al. (2002). In this study no evidence was found for ~~a~~ potential negative effects on the child's behaviour or quality of the parent-child relationship during and after treatment with the Lidcombe Program.

Naturalness of speech in children after treatment with the Lidcombe Program was indirectly investigated by Bonelli et al. (2000) and Onslow et al. (2002). Bonelli et al. (2000) did not observe any change with regards to the parent and child's speech rate, turn-taking behaviour or pragmatics. A number of children in the study developed less quickly than expected in respect to three aspects of language, however development remained borderline normal. Onslow et al. (2002) did not find any evidence of consistent or systematic changes in articulation speed, consonant length, intervocal interval or voice onset time in the 8 boys who stuttered (age 2;7 to 4;11) after treatment with the Lidcombe Program in comparison to the assessment prior to the start of the treatment.

The number of studies into the effect of treatment based on the Demands and Capacities Model compared to the effect of the Lidcombe Program in 2013, during the development of the guideline, was limited to one RCT (Franken et al. 2005). No clinically relevant difference was demonstrated in this study in effectiveness between the Demands and Capacities Model and the Lidcombe Program. The certainty around the effect size is limited. It is very likely that further research will have an important impact on the current estimate of the effect. RCTs into the effect of the Demands and Capacities Model compared to a non-treated control group are not available. This hinders an indirect comparison between the effect of the Demands and Capacities Model and the Lidcombe Program.

The effect on children up to age 6 treated with the Lidcombe Program is large in a short to medium time period (3 to 9 months) in comparison to those not treated. The evidence for this effect is of moderate quality. This implies that there is some uncertainty about the effect size.

Statements about the effect size and the associated degree of certainty in the systematic review and in all RCTs have been based on the outcome measure of stuttering frequency. This outcome measure represents the severity of stuttering, however it is only one aspect determining stuttering severity; the duration of the stutter moment and additional physical concomitant behaviour are other aspects determining the observable severity of stuttering. A very limited amount of indirect evidence has been found for the other outcome measures, such as avoiding speaking situations, naturalness of speech and quality of life. This evidence originates from individual studies each of which investigated the effect of treatment with the

Lidcombe Program. The certainty surrounding the effect size of the reported outcomes is limited.

Conclusions

Certainty of the effect size: Moderate	Treatment of children who stutter up to age 6* with the Lidcombe Program is more effective in the short and medium term (3 to 9 months) in reducing the percentage of stuttered letter groups than when stuttering is not treated (Nye, 2012).

*All children in the studies had had a stutter for at least 6 months at the point of inclusion in order to limit the impact of spontaneous recovery on the result of the experimental research. Conclusions and recommendations about how to treat children who stutter up to age 6 who have been stuttering less than 6 months falls outside of the framework of this report's research question.

Certainty of the effect size: Low	The number of studies into the effect of treatment based on the Demands and Capacities Model is limited and the certainty in respect to the effect size of the DCM is limited. No clinically relevant difference was found in the reduction of the percentage stuttered syllables after treatment between the Demands and Capacities Model group and the Lidcombe Program group.[7] However, there is a great degree of uncertainty about this conclusion given the width of the 95% confidence interval (Franken et al., 2005).

Certainty of the effect size: Very low	There was no evidence found for the potential harmful effects of the Lidcombe Program, such as anxiety, aggression, avoidance and depression. Neither potential harmful effects on the quality of mother and child relationship were found (Woods, 2002).

Speech Motor Training

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[7] De Sonneville, Stolk, Rietveld and Franken (2015) confirmed that no relevant difference was found at 18 months post-treatment onset for percentage of non-stuttering children, stuttering frequency, stuttering severity ratings by parents and therapist, severity ratings by the child, health related quality of life, emotional and behavioural problems and speech attitude. LP and DCM were roughly equal in treating developmental stuttering in ways that surpass expectations of natural recovery.

Speech Motor Training (SMT) (Riley & Riley, 1999) is aimed at improving speech motor planning and thereby reduce stuttering. The programme should not be viewed as a form of therapy in itself, but rather as a supplement to other programmes (Franken & Van Borsel, 2006). The effect of the SMT has been investigated in a single RCT where the reduction in the percentage of stuttered syllables after treatment with SMT was compared with the reduction in the percentage of stuttered syllables after treatment with the Extended Length of Utterance (ELU) (Riley & Ingham, 2000). The age of the children in the study population varied from 3;8 to 8;4 years. SMT reduced the median stuttering percentage by 36.5%. This difference was significant (Wilcoxon matched-pair analysis (z=-2.0, p=0.04)). ELU reduced the stuttering percentage by 63.5%. Mann-Whitney U analysis indicated that the difference in reduction of the stuttering percentage between the SMT and ELU was statistically significant (z= -2.1; p =0 .04).

Riley & Ingham's (2000) study was discussed in two systematic reviews that were included in order to answer the research question (Herder, 2006; Nye, 2012).

Effect size and 95% Confidence interval in the studies with an RCT design in which two interventions are compared without a control group (Nye et al., 2012)

Study	Interventions & Outcome Measures		Statistics				
			Hedges' G	Lower limit CI	Upper limit CI	TX1 n	TX2 n
Riley & Ingham (2000)	SMT vs ELU	%SS	-1.079	-2.209	0.051	6	6

(Detail derived from Table 3, Thesis page 18).

The effect calculated in the systematic review by Nye et al. of these comparative studies was g = -1.079 (95% CI: -2.209 to 0.051). Assuming a threshold value of (-) 0.5 (the threshold value for clinical relevance used by the GRADE working group as a standard) the point estimate of -1.079 indicates a clinically relevant difference in the reduction of the percentage of stuttered letter groups after treatment with SMT compared to treatment based on ELU. However, given the inaccuracy of the result, (the width of the 95% confidence interval is distributed across -2.209 and 0.051 (g = -1.07)), and the high risk of bias (Appendix 1) we are not sure about the effect estimated in this study. The certainty around the effect size is very low.

Conclusion

Certainty of the effect size: Very low	The number of studies investigating the effect of SMT is very limited. Reduction in stuttering percentage after treatment with SMT was significantly lower than reduction after treatment with ELU. However, the clinical relevance of this difference in reducing the percentage is very uncertain given the width of the 95% confidence interval and the high level of risk of bias in the Riley & Ingham study (2000).

Social-Cognitive Behavioural Therapy

Boey's (2003) social-cognitive behavioural (SCBT) therapy in children who stutter is based on social-cognitive learning theory, behavioural therapy, literature regarding cognition and emotion, as well as the general literature on stuttering (Franken & Van Borsel, 2006). The therapy needs to be adapted to the individual needs of the person who stutters or her environment. Boey (2008) describes the central aims of the SCBT as:

- reducing stuttering symptoms;
- preventing and/or limiting the development of stuttering;
- reducing the negative impact in the social domain and on the quality of life.

Specific targets for verbal motor behaviour, the emotional component or the cognitive component of stuttering are formulated depending on the target behaviour. Therapy with children aged up to 6 is carried out in the form of individual sessions combined with a compulsory mentoring programme for parents of 10 to 12 sessions, once every three weeks.

The literature search did not reveal any publications into the effect of SCBT in peer-reviewed journals. No RCTs including SCBT were found. Given that SCBT is often applied in the Netherlands and in particular in Flanders to treat children who stutter, the effectiveness of the SCBT will be discussed in this section using a before and after intervention study which did not include a control group (no treatment or other treatment) (Boey, 2008).[8] The certainty in respect to the effect size in this study is, incidentally, limited given that there was no comparison of two or more homogeneous study groups.

The effect of social-cognitive behavioural therapy has been investigated in the long term and published in a doctoral thesis (Doctor of Medical Sciences, Boey 2008) and presented at various symposia. 122 people were selected at random out of the author's database of 1978 patients who stuttered, of which 87 had received treatment with social-cognitive behavioural therapy, 16 had received a diagnosis of stuttering but had been advised not to undergo

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[8] A control group is defined as a group who in terms of gender, age, initial stuttering severity and potentially other prognostic variables are entirely comparable to the group undergoing the intervention to be evaluated.

therapy and 19 individuals (drop-out group) who refused therapy once the diagnosis of stuttering had been made. The clinical diagnosis was made between 1991 and 2003, and follow-up took place 3 to 14 years, after the clinical diagnosis, through to 2005-2006 with an average follow-up time of 10 years. The outcome measures that were compared related stuttering severity, stutter moments, avoidance, concomitant behaviour, trigger factors, cognitive and emotional symptoms, social reactions and personality characteristics.

Twenty-five children were aged between 1 and 6 during the intake. These children received an average 64 treatment sessions (SD 29.6) and a maximum 121 treatments. The average stuttering percentage in this group during the clinical diagnosis phase was 18.4% Stuttering Like Disfluencies (SLD) (SD 8.7). The average was 0.1% SLD at follow-up (SD 0.3). The difference in reduction on the frequency of stuttering was significant between the group aged 1 to 6 and adolescents ($p < .001$) and between adults and children aged 1 to 6 ($p = .007$), but not in comparison with children aged 7 to 12 ($p = .071$). The initial stuttering severity and the gender (girls) appeared to be important prognostic variables in therapy success.

The certainty in respect to the effect size in the Boey (2008) study is limited due to the design of the study. The final certainty in respect to the effect size after assessment with GRADE is very ~~limited~~ low given the high risk of bias (see Appendix 2).

Conclusion

Certainty of the effect size: Very low	A single intervention study is available with before and after assessments, but no control; this study investigated the effect of Social Cognitive Behavioural Therapy on children who stutter. The average percentage of Stuttering Like Dysfluencies reduced in the 1 to 6 year old age group to 0.1% (SD 0.3). However, there was a great deal of doubt about this outcome given that the study did not have a control group and the high risk of bias (Boey, 2008).

5.4 From Evidence to Recommendations

5.4.1 Certainty with Respect to the Effect Size

The certainty in respect to the degree to which the percentage of stuttering is reduced in children who stutter up to age 6 varies for the different treatment methods. There is a reasonable amount of confidence with respect to the effect size for treatment with the

Lidcombe Program, and the Demands and Capacities Model, whilst there was very limited certainty in respect to the Speech Motor Training and Social Cognitive Behavioural Therapy. The number of studies (RCTs) is limited with the exception of the Lidcombe Program and the studies are small in terms of sample size. In the Franken et al. study (2005) no clinically relevant difference was found in the reduction of the stuttering percentage after treatment between the Demands and Capacities Model group and the Lidcombe Program group. This means it would be premature at the moment to give preference to treatment with the Lidcombe Program instead of the Demands and Capacities Model on the basis of the effects demonstrated.

5.4.2 Balance of Desired and Undesired Effects

The most commonly reported outcome measure is percentage of stuttering, other outcome measures are either not disclosed or rarely reported. The effect on children up to age 6 treated with the Lidcombe Program is large in the short to medium term (3 to 9 months) in comparison to those not treated. There is a greater degree of uncertainty about the effect of treatment with LP in the long term. No undesirable effects of treatment with LP have been demonstrated. The effect of treatment with DCM demonstrated a near identical effect size when compared with LP. The effect size of treatment with SCBT cannot be compared directly with the effect size of LP and DCM given that the effect size of treatment with SCBT was determined using Stuttering Like Dysfluencies (SLDs) instead of the stuttering percentage. The undesirable effects of DCM, SCBT and SMT have not or have barely been investigated or reported.

5.4.3 Values and Preferences

a. **People who stutter**

Given the extent of the effect of treating children using LP for six months or longer after the start of the stutter, compared to not treating, it is important that parents of children who stutter up to the age of 6 are informed about the potential benefits of treating the stutter. Further research may potentially reveal that the effect size for treatment with DCM is comparable, as such the preferences of the parents and the child who stutters may play an important role in determining which of these two treatment methods is the most suitable.

b. **Speech & language therapist / fluency specialists**

With regard to the extent of the effect size for treatment with LP and the degree of certainty it is advisable for practising speech & language therapists and fluency specialists to become skilled in this treatment method and to maintain competencies in LP treatment at a high level. As no clinically relevant difference was found in the effect of treatment with DCM or LP, there

may be a benefit to the practising speech & language therapist or fluency specialist, mastering both treatment methods, to allow a decision to be made in consultation with the parents of the young child who stutters as to which one is to be preferred. Joint decision-making is warranted in that process.

5.4.4 Costs of Therapy

There is hardly information available about the costs of therapy. De Sonneville, Bouwmans, Franken and Stolk (2015) evaluated the incremental cost-effectiveness and cost-utility of the Lidcombe Program compared with treatment based on the Demands and Capacities Model. Differences in effects and costs between the LP and DCM treatment were small. Cost-effectiveness and cost-utility ratios were in favour of the LP. They considered the LP a good alternative to DCM treatment in Dutch primary care. Under Dutch circumstances, effective therapy under both approaches amounts to €2500.

Recommendation

> 11. Discuss with the parents of children up to 6 years of age who stutter the benefits of stuttering treatment no treatment option (reduction of percentage stuttered syllables and stuttering severity). Explain the differences between the 'Lidcombe program' and treatment based on the 'Demands and Capacities Model'. Come to a shared decision with the parents which of the two approaches is preferred.

Rationale for the recommendation

- There is a reasonable degree of certainty that treating children who stutter up to age 6 with the Lidcombe Program is more effective in reducing the percentage of stuttered syllables compared to no treatment.
- Harmful effects of treatment have not been found for the Lidcombe Program.
- There is an increasing degree of trust that the effect of treatment with the Lidcombe Program is no more effective than treatment based on the Demands and Capacities Model and vice versa.
- Differences in effects and costs between the LP and DCM treatment are small. The LP is a good alternative to DCM treatment in Dutch primary care.
- The Lidcombe Program and the Demands and Capacities Model should take preference above treatment with Speech Motor Training and Social Cognitive Behavioural Therapy given the degree of certainty regarding the size of the treatment effect. It should be noted that the Speech Motor Training is not a treatment programme in itself, but a component of a programme. SMT is potentially applicable in treatments based on the Demands and Capacities Model if it appears suitable to the profile of the child who stutters based on the diagnostics.
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Chapter 6: Effectiveness of Stuttering Therapies in Children Aged Between 6 and 13

6.1 Introduction

Children aged between 6 and 13 who stutter will usually have had a stutter for a number of years. The experiences accumulated over the years may lead to anxiety towards the stutter and the associated avoidance behaviours, negative emotions and thoughts that may influence interactions with others. The most commonly employed stuttering therapies for children aged between 6 and 13 in the Netherlands are aimed at the different aspects of stuttering, the emotions and cognitions associated with stuttering and the influence of stuttering on participation in daily life.

The effectiveness of stuttering therapies for children aged between 6 and 13 is discussed in this document. The starting point is that the effectiveness of a stuttering therapy has been evaluated in a randomised controlled trial (RCT) in intervention studies with a control group. Both studies where a stuttering therapy is compared to "doing nothing", as well as studies comparing various stuttering therapies will be considered.

The effectiveness of social-cognitive behavioural therapy (Boey, 2003) will be discussed to supplement the conclusion in order to sketch an up-to-date image of the most commonly used therapies and approaches in the Netherlands.

The draft recommendation for treatment of young children who stutter between age 6 and 13 will be discussed in the last paragraph of this document.

6.2 Method

Three systematic reviews were selected and assessed (Bothe et al., 2006; Herder et al., 2006; Nye et al., 2012) in order to answer the question as to what the desired and undesired effects of stuttering therapies are in children aged 6 to 13 in terms of stuttering severity, avoiding speaking situations, naturalness of speech and quality of life. . The methodological quality of these three systematic reviews is described in the chapter on the desired and undesired effects of stuttering therapy in adolescents and adults, and in the chapter on interventions with young children who stutter.

6.3 Evidence

6.3.1 Overview of the Systematic Reviews of Studies into the Effectiveness of Stuttering Therapies in Children Aged Between 6 and 13

Three different RCTs and two studies with a quasi-experimental design (QED) (Table 1) are discussed in the three systematic reviews included to answer the research question about the wanted and unwanted effects of stuttering therapies in children aged 6 to 13.

Table 1. Summary of the RCTs and QED from the Systematic Reviews by Bothe et al. (2006), Herder et al. (2006) and Nye et al. (2012)

	Design	Intervention	N	Age	Follow-up duration	Outcome measures
Ladouceur & Martineau (1982)	RCT	Breathing regulation, breathing regulation home, control	21 (7/7/7)	5 – 16 yrs.	1 month	Stuttering percentage, Speech rate
Ryan & Ryan (1983)	QED	Modification, DAF[a], Time Out, GILCU[b]	16 (4/4/4/4)	7 – 18 yrs.	6 months	SW/M [c] WS/M[d]
Ryan & Ryan (1995)	RCT	DAF, GILCU	20	7 – 17 yrs.	Could not be defined	SW/M WS/M
Craig et al. (1996)	QED	EMG[e], Intensive Smooth Speech, Smooth Speech Home, Control	97	9 – 14 yrs.	12 months	Stuttering percentage, Speech rate in syllables per minute
Riley & Ingham (2000)	RCT	SMT[f], ELU[g]	12	3.8 – 8.4 yrs.	Could not be defined	Stuttering percentage

[a]Delayed Auditory Feedback, [b] Gradual Increase in Linguistic Complexity of Utterance, [c] Stuttered words per minute, [d] Words spoken per minute, [e] Electromyography Feedback, [f] Speech Motor Training, [g] Extended Length of Utterance

Nye et al (2012)

Nye et al. published a systematic review and two meta-analyses in 2012 evaluating the effectiveness of behavioural interventions on the fluency of speech in children aged between 2 and 18 who stutter. Two randomised controlled trials and a single quasi-experimental study into the effect of stuttering therapies in children aged between 6 and 13 were evaluated in this systematic review. Both RCTs compared the effect of two different interventions against

each other. This concerns a study into the influence of Delayed Auditory Feedback compared to the effect of Gradual Increase in Length and Complexity of Utterance (Ryan & Ryan 1995) and a study where a reduction in the percentage of stuttered letter groups after treatment with Speech Motor Training (SMT) was compared with a reduction in the percentage of stuttered letter groups after treatment with Extended Length of Utterance (ELU) (Riley & Ingham 2000).

The quasi-experimental study by Craig et al. (1996) compared the effectiveness of Electromyographic feedback, Intensive Smooth Speech and Smooth Speech Home with a control that was not treated in terms of the percentage of stuttered syllables and the number of spoken words per minute in children aged between 9 and 14 who stuttered. Participants in this study were not randomly allocated to one of the interventions.

In the systematic review by Nye et al. (2012) the effect calculated for the Craig et al. (1996) study amounted to g = 1.75 (95% CI: 1.05 – 2.45)(Table 2). Assuming a threshold value of (-) 0.5 (the threshold value for clinical relevance used by the GRADE working group as a standard) the point estimate of 1.75 indicates a clinically relevant difference in the reduction of the percentage of stuttered syllables after treatment with Intensive Smooth Speech and EMG. The effect found for both EMG, g = 1.62, as for ISS, g = 2.58, may be considered large. Values greater than 0.7 may be considered as having a large effect (Higgins & Green, 2008). Directly after intervention the percentage of stuttered syllables was, on average, less than 1%. Statistical analyses could not be undertaken in order to maintain data independence due to the comparison of a single control group with three different intervention groups (Nye et al., 2012).

The percentage of stuttered letter groups and the number of spoken letter groups per minute were once again assessed 12 and 52 weeks after the intervention. No statistically significant difference was found (p>0.05) for any of the interventions. The average percentage of stuttered syllables was less than 3% for the three groups. The Smooth Speech Home group and the EMG group scored better than the Intensive Smooth Speech group using a threshold value of 2% for stuttered syllables.

Naturalness of speech was measured pre-intervention, post-interventions and during the follow-up at 12 and 52 weeks after intervention by the researchers, parents and the child on a 5-point Likert scale. Naturalness of speech was assessed as being more natural at the three assessment points after the intervention than before by all of the raters (p<0.001). In comparison with the control group the naturalness of speech in the three treatment groups increased over time according to the parents and researchers. This difference between the treatment groups and the control group was not demonstrated in the assessment of naturalness of speech by the children themselves.

Anxiety was measured with the State-Trait Anxiety Inventory for Children (Spielberger et al., 1972). A significant result was found for time (Wilks' Lambda = .92, df = 4.90, *p*<0.01) for the four different groups directly after the intervention and 12 weeks after the intervention. No significant difference was found between the four groups, including the control group.

The degree of certainty in respect to the effect size was limited due to the quasi-experimental design of the study where the participants were not allocated randomly to a particular intervention or group.

Table 2. Effect size, 95% Confidence interval and p-value for the percentage of stuttered syllables by children aged between 6 and 13 in studies with an intervention versus a control group RCT design and follow-up assessment direct after the intervention (Nye et al., 2012)

Study	Statistics by study					
	Treatment n	Control n	Hedges's *g*	Lower limit	Upper limit	*P* value
Craig et al. (1996)	26	20	1.75	1.05	2.45	<.001

[1] Evaluation using GRADE.

> Risk of bias: reduced by -1 (see Appendix).
> Indirectness: reduced by -1 due to comparison of a single control group with three intervention groups; (To be deducted from the total in the publications given all therapies differ).
> Imprecision: no evidence to warrant a reduction (lower limit of the confidence interval is >0.5);
> Inconsistency: not applicable;
> Publication bias: no evidence to warrant a reduction;
> Report bias: reduced by -1 given not all psychological measures were reported.

Ryan & Ryan (1995) compared the effect of Delayed Auditory Feedback in order to produce slow, delayed speech and thereby reduce stuttering with the effect of Gradual Increase in Length and Complexity of Utterance (GILCU). GILCU is a Fluency Shaping programme based on operant conditioning whereby fluent speech is supported in structured steps. Results were reported for the outcome measures of the percentage of stuttered syllables, stuttered words per minute and the number of words per minute for 20 children aged between 7 and 17, both post-intervention and 14 months after the intervention. The number of stuttered words for 11 participants who completed the programmes reduced from 7.9 to 0.8 in 18.3 hours. Fluent speech was maintained during the assessment 14 months after follow-up.

The systematic review by Nye et al., (2012) calculated the effect in the Ryan & Ryan (1995) study as g = 0.295 (95% CI: -0.797 to 1.387) (Table 3). Assuming a threshold value of (-) 0.5 (the threshold value for clinical relevance used by the GRADE working group as a standard) the point estimate of 0.295 does not indicate a clinically relevant difference in the reduction of the percentage of stuttered syllables after treatment with DAF compared to treatment based on GILCU. The lower limit (-0.797) and upper limit (1.387) of the 95% confidence interval both exceed the threshold value of (-) 0.5 for clinical relevance. This implies the possibility DAF is more effective than the GILCU, or conversely also that the GILCU is more effective than the DAF.

The inaccuracy of the result (the width of the 95% confidence interval spread across -0.797 and 1.387 (g = 0.295)) and the high risk of bias (Appendix A) means we are unsure whether

the estimated effect in this study is close to the actual influence of treatment with DAF or GILCU. The quality of the evidence found was assessed as very low.

Table 3. Effect size and 95% Confidence interval in the studies with an RCT design in which two interventions are compared without a control group (Nye et al., 2012)

Study	Interventions & Outcome measures		Statistics				
			Hedges' *G*	Lower limit CI	Upper limit CI	TX1 n	TX2 n
Ryan & Ryan (1995)	DAF[A] vs GILCU[B]	SWM	0.295	-0.797	1.387	5	6
Riley & Ingham (2000)	SMT[C] vs ELU[D]	%SS	-1.079	-2.209	0.051	6	6

[A]Delayed Auditory Feedback; [B]Gradual Increase in Length and Complexity of Utterance; [C]Speech Motor Training; [D]Extended Length of Utterance;

[1]Evaluation using GRADE. Ryan & Ryan (1995)
> Risk of bias: reduced by -1 (see Appendix A).
> Indirectness: reduced by -1 given the age boundaries of the research population do not correspond with the population in the research question and no subgroup analysis was carried out on 6-13 year olds.
> Imprecision: reduced by -1 (lower limit of the confidence interval is <0.5);
> Inconsistency: not applicable;
> Report bias: reduced by -1 given the absence of relevant information;
> Publication bias: no evidence to reduce scores.

Riley and Ingham (2000) compared the influence of Speech Motor Training (SMT) with the effect of Extended Length of Utterance (ELU) on the outcome measure of the percentage of stuttered syllables in children aged 3.8 and 8.4. SMT is aimed at improving speech motor planning and thereby reduce stuttering. The aim of ELU is to encourage stutter-free speech by offering response-contingent stimulation in meaningful linguistic tasks. SMT reduced the median stuttering percentage by 36.5%. This difference was significant (Wilcoxon matched-pair analysis (z=-2.0, p=0.04)). ELU reduced the stuttering percentage by 63.5%. Mann-Whitney U analysis indicated that the difference in reduction of the stuttering percentage between the SMT and ELU was statistically significant (z= -2.1; p =0 .04).

The systematic review by Nye et al. (2012) calculated the effect of the study into SMT compared with Extended Length of Utterance (ELU) (Riley & Ingham, 2000) as g = -1.079 (95% CI: -2.209 to 0.051) (Table 3). Assuming a threshold value of (-) 0.5 the point estimate of -1.079 indicates a clinically relevant difference in the reduction of the percentage of stuttered syllables after treatment with SMT compared to treatment based on ELU. The inaccuracy of the result (the width of the 95% confidence interval is spread across -2.209 and 0.051 (g = -1.079)) and the high risk of bias (Appendix A) means we are highly doubtful about the effect estimated in this study for children aged 6 to 12, partly as the research population does not fully correspond to the target population in the research question and as no subgroup analysis

has been carried out for this age category. The quality of the evidence found was assessed as very low.

Bothe et al (2006)

The systematic review by Bothe et al. (2006) discusses four studies comparing the effect of stuttering therapy in an RCT or QED in children aged between 6 and 13. This concerns two studies that were also discussed in the systematic review by Nye et al. (2012); Craig et al (1996) and Ryan & Ryan (1995).

The RCT by Ladouceur & Martineau (1982) compared the effect of breathing regulation therapy, with and without the support of parents of children aged between 5 and 15, to a control group that was not treated. The criterion used by Bothe et al. (2006) of less than 5% of stuttered syllables after three weeks of treatment was achieved. No statistically significant difference in the reduction of the stuttered syllables was shown between the intervention group and the non-treated control group after treatment compared to the baseline situation (F= 1.23, *p*>0.05).

Ryan and Ryan (1983) compared the effect of stuttering modification treatment, delayed auditory feedback, pausing and GILCU on the number of stuttered words per minute and the number of spoken words per minute in 16 children who stuttered aged 7 to 18. The authors concluded that fluency of speech increased significantly in the 16 children through all four interventions. The experimental nature of the study, where a non-treated control group is absent, the fact that the number of participants per intervention was very limited and the risk of a very high bias means that we have little confidence in the quality of evidence of this study. The publication does not describe a statistical analysis. This study is not included in the review of evidence in respect to the research question into the effectiveness of stuttering therapy in children aged 6 to 13.

Herder et al (2006)

Herder et al. in their systematic reviews from 2006 discuss two RCTs that evaluated the effectiveness of behavioural interventions on speech fluency of children who stutter aged between 2 and 18. This concerns studies by Ryan & Ryan (1995) and Riley & Ingham (2000). Both studies have been presented in this document in the systematic review by Nye et al. (2012).

6.3.2 Individual (International) Studies into the Effectiveness of Stuttering Therapies in Children Aged Between 6 and 13

This section discusses the study by Hancock et al. (1998) into the long-term follow-up of the Craig et al. (1996) study.

Hancock et al (1998)

The long-term effect of Electromyographic feedback, Intensive Smooth Speech and Smooth Speech Home on the reduction of the percentage of stuttered syllables, spoken syllables per minute, naturalness of speech, degree of recurrence according to parents, the communicative attitude and anxiety were investigated by Hancock et al. (1998). The research population consisted of 62 children who had participated in the Craig et al. (1996) study. The 20 children from the untreated control group were not included in this study given that it is not ethical to deny treatment to children who stutter for 2 to 6 years. The average duration of the follow-up period for the children after randomisation was 4.2 years. The results indicated a potential positive long-term effect of treatment with Electromyographic feedback, Intensive Smooth Speech and Smooth Speech Home; the percentage of stuttered syllables was 75% to 80% lower 2 to 6 years after randomisation than during the assessment time for the intervention. No difference in effectiveness was demonstrated between the treatment groups. The Smooth Speech Home group and the EMG group scored better in the long-term than the Intensive Smooth Speech group when a threshold of 2% for the stuttered syllables was used.

No difference in the assessment of the naturalness of speech was demonstrated in the long-terms in comparison with a 12 months follow-up. Furthermore, no difference was demonstrated between the treatment groups in terms of the naturalness of speech.

The communicative attitude measured using the CAT-R provided an average score of 12.4 (SD = 8.1) for all participants. The average scores for the various treatment groups were virtually the same. The average score of 12.4 was higher than the average for children who did not stutter (M = 8.7), but lower than the average of children who stuttered (M= 16.7) (De Nil & Brutten, 1991).

In terms of trait and state anxiety there were no differences between the various groups and no differences in comparison with the follow-up 12 months after randomisation.

Of the parents 75% indicated that their child spoke fluently during the long-term follow-up assessment; 25% indicated that their child spoke less fluently during this period. 13% of parents had the impression that their child's fluent speech fell back to the level prior to the intervention; 53% had the impression that there was decrease, but not to the level before therapy and 29% indicated that the result obtained through therapy was maintained.

The size of the demonstrated effects was extremely uncertain given that there were no usable follow-up data available for the control group.

6.3.3 Social-Cognitive Behavioural Therapy

Boey's (2003) social-cognitive behavioural therapy in children who stutter (SCBT) is based on social-cognitive learning theory, behavioural therapy, literature regarding cognition and emotion, as well as the general literature on stuttering (Franken & Van Borsel, 2006). The therapy needs to be adapted to the individual needs of the person who stutters or her environment. Boey (2008) describes the central aims of the SCBT as:

- reducing stuttering symptoms;
- preventing and/or limiting the development of stuttering;
- reducing the negative impact in the social domain and on the quality of life.

Specific targets for verbal motor behaviour, the emotional component or the cognitive component of stuttering are formulated depending on the target behaviour.

Therapy with children aged between 7 and 12 is carried out in the form of individual sessions preferably combined with a compulsory mentoring programme for parents of 10 to 12 sessions, once every three weeks.

The literature search did not reveal any publications into the effect of SCBT in peer-reviewed journals. No RCTs including SCBT were found. Given that SCBT is often applied in the Netherlands, and in particular in Flanders, in order to treat children who stutter, the effectiveness of the SCBT will be discussed in this section using a before and after intervention study, that also did not include a control group (no treatment or other treatment) (Boey, 2008).[9] The level of certainty in respect to the effect size in this study is, incidentally, limited given that there was no comparison of two or more homogeneous study groups.

The effect of social-cognitive behavioural therapy has been investigated in the long term and published in a doctoral thesis (Doctor of Medical Sciences, Boey 2008) and presented at various symposia. A random 122 people were selected out of the author's database of 1978 patients who stuttered of which 87 had received treatment with social-cognitive behavioural therapy, 16 had received a diagnosis of stuttering during that time but had been advised not to undergo therapy and 19 individuals (drop-out group) who refused therapy once the diagnosis of stuttering had been made. The clinical diagnosis was made between 1991 and 2003, and follow-up took place 3 to 14 years after the clinical diagnosis in 2005-2006 with an average follow-up time of 10 years. The outcome measures that were compared related to stuttering severity, stuttering moments, avoidance and co-movements, trigger factors, cognitive and emotional symptoms, social reactions and personality characteristics.

Boey (2008) concluded that 32% of the therapy group at the follow-up were still diagnosed as persons who stutter and 55% still reported a stutter. In the diagnosis group 73% were no longer stuttering after correcting or a potential spontaneous recovery. Of the drop-outs 68%

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[9] A control group is defined as a group who in terms of gender, age, initial stuttering severity and potentially other prognostic variables are entirely comparable to the group undergoing the intervention to be evaluated.

were still stuttering during follow-up and 79% reported they were still stuttering. The diagnosis of stuttering was made on the basis of more than 3% Stuttering Like Dysfluencies (SLD) measured in a speech sample of 100 words during a dialogue in the author's clinic.

Twenty-four children were aged between 7 and 12 during the intake. These children received an average of 75.5 treatment sessions (SD 29.3) and a maximum of 140 treatments. The average stuttering percentage in this group during the clinical diagnosis phase was 13.3% SLD (SD 7.1). The average was 1.3% SLD at follow-up (SD 2.2).

The outcome for stuttering severity was measured using the Stuttering Prediction Instrument (SPI) for some children aged between 7 and 12 who stuttered and with the Stuttering Severity Instrument for others. The total scores were converted into z-scores in order to compare the scores from all the participants (the children aged between 7 and 12 who stuttered). The reduction in stuttering severity is more limited in children up to age 6, and is stronger in adolescents and adults. Individual results in terms of the stuttering severity in children aged between 7 and 12 were less homogeneous than in younger children.

Results on the outcome measures of avoidance and co-movements, trigger factors, cognitive and emotional symptoms, social reactions and personality characteristics were not described by age category.

The certainty in respect to the effect size in the Boey (2008) study is limited due to the design of the study. The ultimate certainty in respect to the effect size after assessment with GRADE is very limited given the high risk of bias (see Appendix B).

Conclusion

| Certainty of the effect size:

 Low to very low for all outcome measures | The number of studies into the effects of treatment of children aged between 6 and 13 who stutter is limited and the quality of evidence of the effect is mostly very poor.

 Answering the research question for the age group 6 to 13 is hindered by the fact that the age limits used in the research population in the majority of studies is broader than the 6 to 13 age category and because no analyses have been conducted for this subgroup.

 An exception to this are the studies by Craig et al. (1996) and Hancock et al. (1998) into the effect of Electromyographic feedback, Intensive Smooth Speech, Smooth Speech Home and the study by Boey (2008) into the effect of Social Cognitive Behavioural therapy and stuttering. Electromyographic feedback, Intensive Smooth Speech, Smooth Speech Home and |

	Social Cognitive Behavioural therapy have a positive effect on the stuttering frequency directly after intervention and in the long term, varying from 2 to 14 years after the intervention.
	There is a great degree of doubt about the effect size for these therapies. This is due to the quasi experimental nature of Craig's study (1996), the absence of a control group in Boey's study (2008)[1] and the limited number of participants per research group, meaning less accurate results were obtained.

6.4 From Evidence to Recommendations

6.4.1 Certainty in Respect to the Effect Size

The evidence for all outcome measures for the different stuttering therapies for children aged 6 to 14 is of low to very poor quality. This means that there is a great deal of uncertainty about the treatment effect size resulting in the speech & language therapist / fluency specialist not knowing what effect he/she can expect when implementing treatment for his/her patient and consequently is unable to accurately inform the patient about the treatment effects. This is because of the limited number of RCTs or because these are studies where participants were not randomised into a treatment group or a control group or where a control group was lacking. In terms of the sample size the studies were small and the research population of the RCTs exceeded the age limit of 6 to 13 years. There was also an absence of analyses for the subgroup aged 6 to 13. From this perspective there is no reason to prefer one of the stuttering therapies discussed over the others.

6.4.2 Balance of Desired and Undesired Effects

The effect of treating children aged 6 to 13 who stutter with Electromyographic feedback, Intensive Smooth Speech, and Smooth Speech Home on the percentage of stuttered syllables is large immediately after the intervention when compared to no treatment. A positive effect is demonstrated 12 months and 2 to 6 years after treatment. A positive influence was also demonstrated in the short and long term in respect to the perceived naturalness of speech. No differences were demonstrated in respect to trait and state anxiety between the various groups and the untreated control group. An average reduction of 90.2% in the percentage of Stuttering Like Dysfluencies was reported in the Boey (2008) study compared to the percentage before the intervention. The reduction in stuttering severity is more limited in children up to age 6, and is stronger in adolescents and adults.

The effect sizes for both studies cannot be directly compared with each other due to the fact that different outcome measures were used in the studies. There is a great deal of uncertainty about the effect described in the Craig et al. (1996), Hancock et al. 91998) and Boey (2008) studies.

6.4.3 Values and Preferences

a. People who stutter

As there is an absence of a reasonable, or even a large degree of certainty about the effect size of the different stuttering therapies for children aged 6 to 13 for all outcome measures, the preferences of children who stutter and their parents may play an important role. It is important that parents of children aged 6 to 13 who stutter and the children themselves are informed about the potential benefit of treating the stutter. There are indications that the treatment of stuttering between the age of 6 and 13 can significantly reduce the severity of the stutter compared to treating the stutter during adolescents or in adulthood. Given there is insufficient evidence that one stuttering therapy is more effective than another, parents and children who stutter should, in consultation with a speech & language therapist or fluency specialist, decide which treatment method is preferred.

b. Speech & language therapist - fluency specialist

See text for adults.

6.4.4 Costs of Therapy

No data are available regarding the costs incurred in the treatment of children who stutter aged 6 to 13. For this population it is therefore also desirable that evidence around cost-effectiveness is collected.

Recommendation

12.	The treatment of stuttering in children between the ages of six and thirteen years should be based on a treatment plan that contains all ICF elements and focuses on the types of behaviors, emotions and cognitions that have been identified, in collaboration with the child who stutters and his parents during assessment.

Rationale for the recommendation

- There is a (very) limited degree of certainty regarding the extent to which the number of stuttered syllables is reduced in children aged 6 to 13 after treatment.
- There is insufficient evidence that one stuttering therapy is more effective than another in the treatment of children who stutter aged 6 to 13.

- There is some but limited degree of certainty that a reduction in stuttering severity in children aged 6 to 13 is less than in children up to age 6, but stronger in adolescents and adults.
- Joint decision making concerning the objectives on the basis of the diagnostics and in consultation with the child who stutters and the parents is warranted given the multifactorial nature of stuttering.

Recommendations for further research

Given the lack of high quality evidence for the effectiveness of stuttering therapy in children aged 6 to 13, further research is recommended into the effectiveness of stuttering therapy in this target group.

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Chapter 7: Effectiveness of Stuttering Therapies in Adolescents and Adults

7.1 Introduction

The effectiveness of various stuttering therapies for adolescents and adults will be described in this chapter. The starting point is that the effectiveness of a stuttering therapy has been evaluated in comparative research in a "randomised controlled trial" (RCT). Both studies where a stuttering therapy has been compared to "doing nothing", as well as studies where different stuttering therapies have been compared with each other will be considered. The success rate of stuttering therapies will be evaluated for the following outcome measures: stuttering severity, avoidance behaviours, participation and quality of life, and naturalness of speech. All outcome measures are crucial from the perspective of a person who stutters (PWS), however the last item is of less relevance. As these outcome measures are rarely used all together in the various studies a very strict comparison is often difficult.

Viewed from an historical perspective, two streams may be distinguished within stuttering therapy: "stuttering modification therapy" and "fluency shaping therapy". The aim of "stuttering modification therapy" is to be able to stutter more freely and easily. The reduction in anxiety for stuttering is the central starting point. In "fluency shaping therapy" the stuttering itself is the central point and the person who stutters is taught through a behavioural therapy programme to speak more fluently step by step (Bezemer et al., 2010).

However, in practise and in scientific research a diversity of methods, working models and treatment techniques are available of which "stuttering modification therapy" and "fluency shaping therapy" may be incorporated into. Multiple therapies and therapeutic techniques will also be considered in this text.

7.2 Method

A search was undertaken on 30 July 2012 for systematic reviews (with or without meta-analyses) of RCTs. The following sources were used for this with the potentially relevant systematic reviews and the total studies found included in brackets: PsycInfo (0/29), Embase (0/11), Cinahl (0/37), Cochrane Library (0/13) and Medline (6/111). Of the five studies and the potentially relevant systematic reviews found in Medline, one appeared to be a commentary on a systematic review. The criteria for determining the relevance were:

- Subject pertained to stuttering

- Study participants were adolescents or adults
- The study pertained to a therapeutic (stuttering therapy or pharmacotherapy) and not a diagnostic intervention.[10]

A search was undertaken on 12 February 2013 using the above sources for original RCTS, including an update of the systematic reviews; the number of potentially relevant RCTs and total number of studies found is included in brackets: Embase (4/94), Cinahl (4/108), Medline (7/99) and PsycInfo (3/107).

The criteria for determining the relevance were the same as before, with in addition that the potentially relevant RCTs that were found had not already been included in a systematic review.

Five relevant systematic reviews and eight relevant RCTs were found for both searches. These did not include any research from the Netherlands. For this reason a discussion of the study by Huinck and Peters (2004) has also been added. In this longitudinal study three methods applied in the Netherlands were evaluated.

For details of the search strategy please refer to Appendix 5.

7.3 Evidence

The various systematic reviews will be discussed in section 3.1. The individual studies which have been subsequently published or not included in the reviews will be discussed in section 3.2. The therapies evaluated in the Netherlands will be discussed in section 3.3.

7.3.1 Overview of Systematic Reviews into the Effectiveness of Behavioural Interventions

Bothe et al. (2006)

In 2006, Bothe et al. published a systematic review of studies published between 1970 and 2005, which evaluated behavioural interventions, cognitive interventions and related interventions for stuttering. There are a number of criticisms in terms of how this review was carried out. For instance, the description of the interventions and the composition of the study groups in terms of sex, age and presence of co-morbidity were not described in much detail. In addition to this, the level and figures were not provided in a tabular format for the outcomes, meaning that a comparison between and within types of interventions is difficult.

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[10] The effectiveness and side-effects of pharmacotherapy have been described elsewhere.

Furthermore, the reviewers also used a measure of success for frequency of stuttering - less than 5% stuttered syllables after intervention - which does not lend itself easily to discussion (see also Appendix 3).

Virtually all studies evaluated an intervention falling under the term "fluency shaping therapy". In terms of adults the majority of studies described treatment of the type "prolonged/smooth speech" and "regulated breathing and airflow".

Experimental, comparative studies with randomisation (intervention A versus intervention B or intervention A versus doing nothing/placebo) are the best evaluation methods for assessing the positive and negative effects of interventions. Follow-up outcomes (6 months, 12 months after the end of treatment) are much more relevant for the person who stutters than outcomes at the end of treatment. There were two studies with a comparative design, but no randomisation.[11] These evaluated "prolonged/smooth speech" in adults.[12] These two studies also presented follow-up details per study (Table 1). The follow-up duration was 6 months. The frequency of stuttering at the end of treatment and at the end of follow-up had reduced by approximately 50-80%. It is noticeable that the frequency of stuttering did (gradually) increase again during the follow-up period. In terms of speech tempo there appeared to be some increase as a result of the therapy. James et al. (1989) also investigated to what extent social, emotional or cognitive variables improved. Their study group who followed DAF and therapy breathing frequency, phrasing and prosody indicated that they did not find it as problematic to talk and therefore did not avoid talking as much.[13]

Table 1. Frequency of stuttering and speech rate after various "prolonged/smooth speech" therapies

		Frequency of stuttering (% stuttered syllables)				Speech rate (number of syllables per minute)			
		Baseline	PT*	FU*	% improvement (FU-baseline)	Baseline	PT*	FU*	% improvement (FU-baseline)
Perkins 1974									
N=44	Fluency training								

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[11] A decision was made to only determine the risk of bias and the certainty of the effect size for those studies reporting the confidence intervals. In the absence of confidence intervals it is not possible to arrive at a judgment about the accuracy of the effect size.
[12] This relates to studies the reviewers have coded as "multiple groups, no random assignment"; "multiple groups, random assignment".
[13] In the Perkins et al. study no information was provided about the level of reduction.

		Frequency of stuttering (% stuttered syllables)				Speech rate (number of syllables per minute)		
m/f: 37/7	-CF** condition	7.82	2.41	2.98	62	163	193	18
Age: 12-52 (virtually only adults	-TO** condition	5.17	1.75	1.67	68	175	189	8

James 1989

N=24 (4 dropouts) m/f:18/2	Delayed auditory feedback (DAF)	16.32	2.64	8.44	48	230	-	245	6
Age:19-50	DAF and therapy for breathing, phrasing and prosody	9.04	1.04	1.73	81	245	-	249	2

Median percentage improvement: 65 (range: 48-81%)   Median percentage improvement: 7 (range: 2-18%)

* PT: post treatment; FU: follow-up period of ≥ 6 months. ** CF: "contingency free"; TO: "time –out"

Bothe et al. (2006) reported on two studies using regulated breathing and airflow where the frequency of stuttering was gauged during follow-up. The follow-up period for these studies was 6 or 27 months. Favourable effects on the frequency of stuttering were also seen for these forms of therapy (Table 2). The degree of improvement varied from 35-67% less for stuttered syllables. These outcomes were slightly less positive than those for "prolonged/smooth speech". This did not appear to apply for speech rate: breathing therapy seemed to be somewhat more beneficial. However, in both cases there was considerable variation within these forms of therapy (Table 1 and Table 2).

Table 2. Frequency of stuttering and speech tempo after various "regulated breathing and airflow" therapies (with potentially additional therapies)

		Frequency of stuttering (% stuttered syllables				Speech rate(number of letter groups per minute)			
		Baseline	*PT**	*FU**	*<t score="N/A">*	*Baseline*	*PT**	*FU**	*% improvement (FU-baseline)*
Ladouceur 1986									
N=16 m/f:12/4 Lft: 18-37	regulated breathing method & systematic desensitisation & cognitive restructuring								
	-"Persons who stutter"	7.7	3.7	2.5	67	No change in respect to "persons who do not stutter"			
	-"Persons who do not stutter"	2.5	-	1.8	28				
Saint-Laurant 1987									
N=40 m/f: 27/13 Lft: 18-50	- Intensive regulated-breathing	9.3	4.1	5.4	42	186	204	224	20
	- Spaced regulated-breathing	9.2	4.7	4.7	49	163	190	212	30
	- Intensive regulated-breathing and maintenance phase	9.3	4.2	5.0	46	171	186	204	19
	- Spaced regulated-breathing and maintenance phase self-	10.8	5.4	7.0	35	153	175	178	16

	Frequency of stuttering (% stuttered syllables)				Speech rate(number of letter groups per minute)			
	Baseline	PT*	FU*	<t score="N/A">	Baseline	PT*	FU*	% improvement (FU-baseline)
recording procedure (=placebo)	9.8	8.4	-	-	145	156	-	
	Median percentage improvement: 44 (range: 35-67%)				Median percentage improvement: 19 (range: 16-30%)			

* all values were graphically interpolated

Ladoucer and Saint-Laurent (1986) investigated whether communicative attitude (using the Erickson scale of communication attitudes) and the presence of irrational beliefs in persons who stutter change through breathing therapy in combination with desensitisation and cognitive restructuring. They were unable to ascertain any significant change.

Two of the four studies involving breathing therapy without follow-up information showed a reduction in the frequency of stuttering of less than 5%. Other outcome measures were not reported by the reviewers.

Bothe et al. (2006) reported two studies pertaining to adolescents and adults which discussed other types of interventions than those considered here. Öst et al. (1976) compared (1) speech training with a metronome (n=5) and (2) "shadowing" (n=5) with a waiting list control group (n=5). The average age of the trial participants was 25 years (11 men and 4 women). Speech training with a metronome resulted in a statistically significant reduction in the percentage of "non-fluency" from 16.6 to 9.3 after 14 months follow-up, in other words a drop of 44%.[14] "Shadowing" was associated with a non-significant reduction from 14% to 9.6% after 14 months follow-up, or a reduction of 33%. The corresponding figures for the waiting list control group were: 17.2% versus 9.5%, or a drop of 45%.[15] Speech training with a metronome did not alter the number of words read aloud per minute (108 versus 105 at follow-up); nor was any change observed in the waiting list control group. In the "shadowing" group, however, the number of words increased significantly from 75 to 90 per minute.

The Ingham & Andrews (1973) study evaluated a therapy based on rewarding less stuttering with tokens in combination with utilising "delayed auditory feedback". Only the outcomes immediately after treatment were available for this treatment, and no statistical analysis was applied. The diverse graphics in this publication revealed a reduction in the percentage of

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[14] This study did not provide any information about the naturalness of speech.
[15] The 45% reduction was not statistically significant due to the large variation in response within the waiting list control group.

stuttered letter groups from approximately 15.5 to 11 (experiment 1) or 8 (experiment 2, where the reward was doubled). The corresponding figures for the number of spoken letter groups per minute were 80 versus 125 and 110 versus 135.

The review did not investigate to what extent gender, socio-economic status, educational level, age and ethnicity could have influenced the impact of therapy.

An evidence profile was produced (Appendix 3) in order to establish the degree of certainty in respect to the effect size for stuttering therapy.[16] This was categorised as limited due to important limitations in the design and implementation of the studies and due to the considerable heterogeneity in the outcomes. A significant downside to the studies reporting follow-up details which were included in this review is that in only one instance the follow-up period was longer than 24 months.

The reviewers concluded that in adults "prolonged/smooth speech" is effective: "*For adults who stutter, many of the articles that met this review's trial quality criterion support the use of prolonged-speech-type procedures within a comprehensive treatment framework that includes initial intensive work, practice in front of groups, specific transfer or generalisation tasks, self-evaluation of speech and/or self-management of program steps, a focus on speech naturalness and feedback of naturalness measurements, and an active contingent maintenance program that continues to address not only stuttering but also speech naturalness and self-evaluation skills*".

This conclusion does not completely correspond to the conclusion we drew earlier: "prolonged/smooth speech" is potentially more effective in terms of the reduction of the frequency of stuttering, whilst breathing therapy appears more favourable for speech rate. However the latter is less important as an outcome measure. It is possible that the fact that we have focused on studies with a follow-up of at least six months and not so much on outcomes directly after therapy may have led to a different judgment than that of Bothe et al. (2006).

Herder et al. (2006)

Herder et al.'s (2006) systematic review carried out a number of meta-analyses of RCTs where a form of behavioural therapy with operant methods ("behavioral stuttering intervention") was compared with a control group or a different therapy. The implementation of this systematic review was moderate, particularly as none of these studies were assessed in terms of methodological quality (Appendix 2). Six studies were included in the first meta-analysis

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[16] The certainty of the effect size is defined in GRADE as follows.
High: Further research is very unlikely to change our confidence in the estimate of effect.
Moderate: Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.
Low: Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.
Very low: We are very uncertain about the estimate.

where stuttering therapy was compared to a control. Some studies involved adolescents and adults, whilst others focused on children. The combined effect size was 0.91 (95% CI: 0.57 - 1.26) immediately after treatment; values above 0.7 are generally considered as a large effect size.[17] However there was a significant amount of variation in effect sizes in the six studies varying from 0 to 1.87 (Figure 1). We judge the certainty around the effect size to be narrow.[18] The reviewers found four studies in which different types of behavioural therapy were compared in adults and adolescents (Figure 2). The reviewers concluded on the basis of a meta-analysis that there was no difference in the capabilities between the different types of behavioural therapy. An effect size (Hedge's G) of 0.29 does not indicate a relevant difference in effectiveness (Figure 1), given the usual threshold value employed is 0.5.[19] However, the result of the meta-analysis on which this conclusion is based is uncertain, given the fact that the upper limit of the confidence interval (0.76) lies above the 0.5 threshold value. The certainty around the effect size may be categorised as limited.[20]

Figure 1. Studies where the effectiveness of different behavioural therapies for children, adolescents and adults were compared against a control group in RCTs

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[17] Holger J Schünemann, Andrew D Oxman, Gunn E Vist, Julian PT Higgins, Jonathan J Deeks, Paul Glasziou and Gordon H Guyatt on behalf of the Cochrane Applicability and Recommendations Methods Group (2008). Chapter 12 Interpreting results and drawing conclusions. Published by John Wiley & Sons, Ltd under "The Cochrane Book Series" Imprint.

[18] Assessment with GRADE.
Risk of bias: not assessed;
Indirectness: reduced by -1 as children and adults were not analysed separately;
Imprecision: no evidence to warrant a reduction (lower limit of the confidence interval is >0.5);
Inconsistency: reduced by -1. Although the p-value for the heterogeneity test was not significant (p=0.13), the I^2 was 41.07 and the point estimates varied considerably.
Publication bias: no evidence to warrant a reduction.

[19] The 0.5 value should be read as half a standard deviation. In other words if the result of two interventions differs by more than half a standard deviation, this difference will, in principle, be classified as a relevant (clinical) difference.

[20] Assessment with GRADE.
Risk of bias: not assessed;
Indirectness: no evidence to warrant reducing the value;
Imprecision: reduced by -1 as 0.5 falls within the confidence interval (-0.18; 0.76);
Inconsistency: reduced by -1. Although the p-value for the heterogeneity test was not significant, the point estimates (-0.01,; 0.68; 0.22; 030) differed considerably;
Publication bias: no evidence to warrant a reduction.
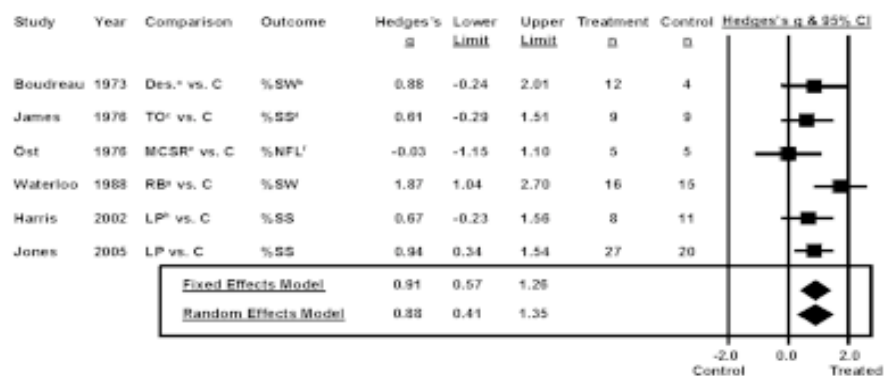
Source: Herder et al. (2006)

Figure 2. Studies where the effectiveness of different behavioural therapies for adolescents and adults were compared against a control group in RCTs
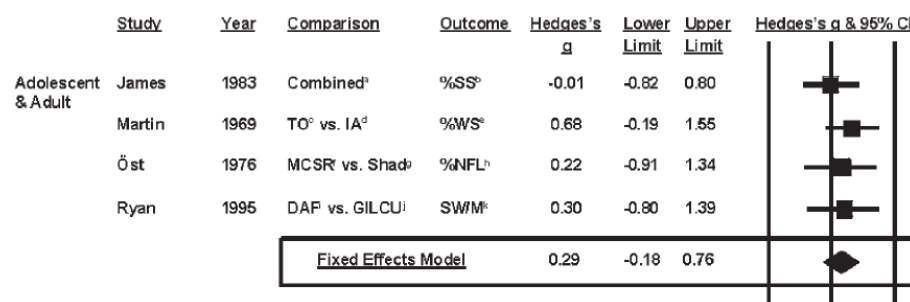
Heterogeneity statistics for a fixed model: Q=4.38, df=6, p=.63, I Squared=0.00

[a] Experimenter-Administered Time-Out, Self-Initiated Time-Out, and Experimenter-Administered Self-Terminated Time-Out; [b] Percent Stuttered Syllables; [c] Time Out from Speaking; [d] Information-Attitude; [e] Percent Words Stuttered; [f] Metronome-Conditioned Speech Retraining; [g] Shadowing; [h] Percent Non-Fluency; [i] Delayed Auditory Feedback; [j] Gradual Increase in Length and Complexity of Utterance; [k] Stuttered Words per Minute; [l] Lidcombe Program; [m] Demands and Capacities Model; [n] Lidcombe Program with and with out Parental Verbal Contingencies for Stuttering and Stuttering Severity Ratings; [o] Speech Motor Training; [p] Extended Length of Utterance

Source: Herder et al. (2006)

Bate et al. (2011) and Woodman & Moore (2011)

The aim of the review by Bate et al. (2011) was to investigate the influence of the so-called "habit reversal" therapies for tics, habit disorders and stuttering. Two studies were included in this review pertaining to stuttering therapy in adults. Both studies involved an evaluation of the effectiveness of breathing therapy and were included in the Bothe et al. (2006) review. The aim of the Woodman & More (2011) review was a systematic evaluation of the scientific literature concerning the effectiveness and safety of applying the Alexander technique for health-related conditions. The reviewers localised the Alexander technique in the field of "complementary medicine and alternative approaches". They reported a single study which evaluated the application of the Alexander technique in 30 one-on-one lessons with two trial participants. Eight of the 17 non-validated physical and psychological outcome measures demonstrated a non-quantified, statistically significant improvement (p<0.01 and p<0.05).

According to the reviewers there was "preliminary evidence suggesting improvement in stuttering, but this evidence is insufficient to support recommendations in this area".

7.3.2 Individual (international) Studies into the Effectiveness of Cognitive and/or Behavioural Interventions appearing after the Publication of the Systematic Reviews

Carey et al. (2010) carried out an RCT (n=40, of which 17.5% female) in a university setting where a telehealth version of the Camperdown programme – a programme for restructuring speech with a focus on breathing, phonation and articulation - was compared with the regular face-to-face programme.[21] The researchers used the following exclusion criteria: younger than 18, a frequency of stuttering less than 2%, insufficient knowledge of English and treatment for stuttering received during the previous 12 months. The programme has four components: individual learning sessions, a group day in order to practise, individual sessions aimed at solving problems and maintenance/support of the learned fluency techniques. The telehealth version was adapted as follows: the entire treatment was undertaken by phone; prolonged speech and naturalness of speech were taught through audiotapes sent to study participants; a specific voice mail line could be used to record speech samples; instruction and feedback were provided as required by the fluency specialist; home exercises replaced the group day in the face-to-face programme. The primary outcome measures were percentage of stuttering and contact time; secondary outcome measures were naturalness of speech (evaluated with a standardised instrument), self-reported stuttering severity (using a 9-point scale) and treatment satisfaction.[22] The follow-up was after 12 months. This was a well-implemented study with a low risk of bias (see Appendix B). The degree of certainty regarding the effect size is limited.[23]

In terms of the difference in the percentage of stuttering between both groups, no significant difference (p=0.9) was found immediately after treatment nor during the 6 or 12 months follow-up. The average percentage of stuttering was 2.4 (immediately after treatment), 2.8 (6

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[21] No RCTs were found in which the standard form of the Campderdown Program had been investigated as such in terms of effectiveness.

[22] The percentage of stuttering was determined by an indepedent speech & language therapist using a 10 minute audio recording of a telephone conversation of the person who stuttered with three others; two strangers and the therapist.

[23]Assessment with GRADE. For both outcomes:

Risk of bias: no evidence to warrant a reduction;

Indirectness: no evidence to warrant a reduction;

Imprecision (-1): the confidence intervals for these two outcome measures were absent, therefore the level of accuracy was difficult to determine;

Inconsistency: not applicable;

Publication bias (-1): a stage 3 RCT was undertaken to evaluate the effectiveness of the standard version of the Camperdown Program. For instance, O'Brian et al. (2003) have written: "The promise of this Stage 2 clinical trial has led the authors to initiate a Stage 3 randomised controlled trial of the Camperdown Program". However, no publication could be found. Therefore there may be an element of publication bias.

months after treatment), and 2.6 (12 months after treatment). The percentage of stuttering was estimated at 7 in the telehealth group at the start of therapy, and roughly 5.4 in the face-to-face group; this difference was corrected in the analysis. The reduction in the percentage of stuttering was therefore approximately 60%. Self-reported stuttering severity (1 = not stuttering, 9 = very severe) was reduced in the telehealth group from 3.9 to 2.3 after 9 months, and from 3.8 to 2.4 in the face-to-face group, a reduction of relatively 40%. The authors did not report any figures in terms of the naturalness, although they did only note that there was no difference between telehealth and the face-to-face group. The telehealth version was more frequently described as "particularly appropriate" (p=0.018). The table below provides an overview of the effect of various variables on the contact time with the fluency specialist.

Table 3. Difference in contact time (minutes) with a fluency specialist between the treatment groups corrected for all other variables

Variable	Estimate	95% confidence interval
Telehealth version	-221	-387; -56
Women	227	0; 453
Older participants	-15	-25; -4
Stuttering severity before treatment	213	41; 384
Previous treatment	361	171; 551
Family history	17	-152; 185

Cream et al. (2010) carried out a multicentre RCT at a university setting with 89 adolescents and adults who stuttered (m:f=4:1; average age: 27 years) in which restructuring speech with standard maintenance was compared with the same intervention supplemented with video self-modelling. Video self-modelling was defined by the researchers as follows: "a behavioural intervention during which people view video images of themselves free of a problem target behaviour". The following exclusion criteria were used: less than 2 on the percentage of stuttering; insufficient command of English; stuttering that started after the age of 12; stuttering resulting from a psychological disorder; treatment in the previous 6 months. Three centres applied the "La Trobe smooth speech programme" and three centres a modified version of the Camperdown programme. According to the researchers both programmes had previously shown the same outcomes; the percentage of stuttering was the primary outcome

measure.[24] There were 5 secondary outcome measures: self-reported anxiety, self-reported stuttering severity, avoiding speaking situations, quality of life and satisfaction with fluent speaking. The follow-up amounted to 6 months (calculated from the point of randomisation). This concerned a well-conducted study with a low to moderate risk of bias (see Appendix B), although compliance (watching the video daily) was only 50%, while almost 40% did not provide any information about the frequency of viewing. In terms of the difference in the percentage of stuttering the video self-modelling did not add anything: at the end of the follow-up period the difference in the percentage of stuttering was 0.06 (95% CI: –1.3 to +1.4 stuttering percentage). For the secondary outcome measures a statistically significant difference was seen on some, but not on others, and therefore it is unclear if these were clinically relevant differences. There was a great degree of certainty in respect to the effect size for the outcome percentage of stuttering and limited for the secondary outcome measures.[25]

Lincoln et al. (2010) investigated in 11 adults (age: 21-65 years; m:f=7:4) whether four different combinations of auditory feedback – (minimum/maximum) delayed auditory feedback (DAF), (minimum/maximum) frequency-altered feedback (FAF) and masking auditory feedback (MAF) – have a different effect during conversation on the percentage of stuttered syllables in comparison to normal auditory feedback.[26] An important limitation of this study is that it was not clear to what extent the *sequence* in which the combinations were evaluated affected the results. The results of this study have been summarised in Table 4. There were no statistically significant differences between the control condition and the other conditions. The very wide confidence intervals illustrate this. This may possibly be traced back to a great deal of inter-individual variation on the one hand and a small sample size on the other. The certainty in terms of the effect size is restricted.[27] The researchers concluded that:

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[24] Study percentage was determined using two 10 minute audio recordings of telephone conversations with unknown research assistants who recorded the conversations.

[25]Assessment with GRADE. There was no evidence for the outcome percentage of stuttering to reduce one of the GRADE factors. However that was not the case for the other outcome measures:

Risk of bias: no evidence to warrant a reduction;

Indirectness: no evidence to warrant a reduction;

Imprecision: no evidence to warrant a reduction;

Inconsistency (-2): no consistency for quality of life and other outcome measures;

Publication bias: no evidence to warrant a reduction.

[26]Percentage of stuttering was determined using video recordings by a speech & language therapist experienced in assessing stuttering and unaware of the aim of the study and the circumstances in which the conversations were being held. In order to do this he watched the DVDs with speech samples and scored the percentage of stuttering in real time with the help of a handhold tool.

[27] Risk of bias (-1): implementation of the study not entirely clear (see text);

Indirectness: no evidence to warrant a reduction;

Imprecision (-1): very wide confidence intervals;

Inconsistency: no evidence to warrant a reduction;

Publication bias: no evidence to warrant a reduction.

"Participants' varying responses to differing Altered Auditory Feedback (AAF) settings likely accounted for the failure to find group differences between conditions. These results suggest that studies that use standard DAF and FAF settings for all participants are likely to underestimate any AAF effect. It is not yet possible to predict who will benefit from AAF devices in everyday situations and the extent of those benefits".

Table 4. Percentage difference in stuttering percentage of various combinations of altered auditory feedback with normal auditory feedback

Condition/Combination	Stuttering percentage difference	95% confidence interval
1. Control (NAF)	— —	
2. MIN FAF + MIN DAF	−27	−65; 53
3. MAX FAF + MAX DAF	−49	−76; 8
4. MAX FAF + MIN DAF	−31	−67; 44
5. MIN FAF + MAX DAF	−39	−71; 28
6. MAX FAF + MAX DAF (READING)	−62	−90; 40
7. White noise (MAF)	−51	−77; 3
8. Control reading (NAF)	— —	

Hewat et al. (2006) investigated in 22 adolescents and adults whether behavioural therapy based on learning to pause after a moment of stuttering (self-imposed time-out) is effective. They used the following inclusion criteria for this: age younger than 14, sufficient command of English, no stuttering therapy in the previous 12 months. This study was set-up on the grounds of the following principles, amongst others: '(…) *unlike prolonged speech, which must be used continually to suppress stuttering, time-out is only applied at times of stuttering. Consequently, it is likely to be less socially conspicuous than a novel speech pattern (…) Time-out appears to be a flexible means to control stuttering because its viability does not depend critically on either the duration of the time-out period or the consistency of the application of the stimulus (…). Time-out would appear to be capable of producing clinically significant reductions in stuttering in far fewer treatment hours than prolonged-speech*". In terms of the last point, the researchers needed to establish on the basis of their results that restructuring speech provides a more predictable clinical response and is associated with a stronger reduction in the percentage of stuttered syllables than the time-out approach. Furthermore

according to Hewit et al. (2006) it was also necessary "*to identify those clients who are most likely to respond to the program*".[28]

Menzies et al. (2008) employed an RCT (N=32; 2 dropouts; m:f = 25:5; age: 18-66) to investigate if cognitive behavioural therapy (CBT) plus stuttering therapy (speech restructuring) could reduce social anxiety and stuttering (more than) stuttering therapy (speech restructuring). The exclusion criteria were: treatment with cognitive behaviour therapy in the previous 6 months' or speech treatment in the previous 12 months; inadequate command of English; learning disability; transient suicidal ideation; current use of benzodiazepines; not willing or able to maintain a stable dose of psychotropic medication during the study. The CBT programme consisted of 10 weekly sessions of individual treatment amounting to a total of 15 hours, and was therefore specifically aimed at reducing speech-related anxiety. This programme was presented prior to stuttering therapy for the experimental group, whilst the control group did not receive an intervention in that period. The follow-up duration was for 12 months.

At the start of the study, 67% of the experimental group and 53% of the control had a diagnosis of social phobia. After 12 months there was no one in the experimental group with this clinical diagnosis. The diagnosis had been made by a psychologist who was not aware to which study group the participants had been assigned. The percentage of individuals with a social phobia in the control group remained unchanged. The results were slightly under statistical significance due to the small study sample size (Fisher's exact test, p = 0.0055, odds ratio 2.0, [95% CI: 0.9 – 4.5]. The CBT programme appeared to have a substantial, positive effect on general functioning as measured by the Global Assessment of Functioning scale (GAF scale with scores between 0 to 100). After following the CBT programme the score of the GAF scale was 21 points (95% CI: 12.6-32.7) higher in the control group. This means that those who had followed the CBT programme were able to carry out more everyday tasks with fewer psychiatric difficulties, less anxiety and less avoidance behaviours. Following the CBT programme probably also led to more activities high in the "anxiety hierarchy" being carried out: 33% (p<0.005) more than in the control group. The scores on the Unhelpful Thoughts and Beliefs About Stuttering (UTBAS) instrument also demonstrated a significant drop at various time-points, however at the end of the follow-up these were no longer significant. At the start of the study, the percentage of stuttering in the control group was approximately 8.5 and estimated at 7 in the experimental group.[29] At the start of the stuttering therapy the percentage of stuttering had reduced by 11% in both groups. Directly after treatment the

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[28]This study was non-comparative in design, and can therefore only be characterised as evidence providing a limited degree of certainty in respect to the effect size. A comparative design was, in principle, easily achievable. In that design a control group could have followed behavioural therapy based on speech restructuring and the experimental group could have followed behavioural therapy based on time-out.
[29]Stuttering percentage was determined using two recordings of 10 minute conversations with administrative staff or students at the clinic. This was undertaken by a speech & language therapist experienced in assessing stuttering, but unfamiliar with the aim of the study.

reduction in the percentage of stuttering was 35% and 70% in the experimental and control group respectively, *insofar as were these involved people diagnosed with a social phobia.* After 12 months these percentages were 56% and 88% in the experimental and control group respectively with otherwise no statistical significance. The certainty around the effect size was limited for the outcome measures "UTBAS" and social phobia, and moderate for "functioning".[30]

Iverach et al. (2009) investigated whether the presence of psychiatric disorders in people who stutter contributes to the inability to maintain fluent speech after treatment. They recruited 64 adults (m:f=4:1, average age: 32 years) for a study which evaluated the effect of self-modelling on maintaining the benefits of speech modification. The average percentage of stuttered syllables at the start of treatment was 8.3. The CIDI-Auto-2.1 (**C**omputerised Version of the Composite **I**nternational **D**iagnostic **I**nterview*)* – a standardised computer interview which has been shown to be reliable and valid for scientific research - was used to determine the psychiatric problems. In addition to this the IPDEQ (**I**nternational **P**ersonality **D**isorder **E**xamination **Q**uestionnaire) was used to screen for nine personality disorders (paranoidal, schizoidal, antisocial, impulsive, borderline, histrionic, obsessive-compulsive, anxiety disorder and dependent personality disorder). The researchers investigated for various variables (a personality disorder, an anxiety disorder, a mood disorder, presence of 1-2 psychiatric disorders, more than 3 psychiatric disorders) at *three timepoints* (prior to treatment, immediately after treatment, and 6 months after treatment) to observe whether there was a difference in the percentage of stutteredsyllables, self-reported stuttering severity and self-reported avoidance behaviour.[31] This resulted in carrying out a statistical test 5 * 3 = 15 times per outcome measure, which increased the risk of a chance finding. The more obvious approach - given the study's research question - would have been to analyse whether, for instance, the difference in the percentage of stuttered syllables *was the same or different after 6 months follow-up* for those who did not have a personality disorder and those who did have a personality disorder. Either way, the researchers did not find a statistically significant difference in the main. However, they did find an association for the outcome measure percentage of stuttered syllables after 6 months: individuals with 1-2 psychiatric disorders stuttered more in comparison with those who did not have a psychiatric disorder. For the outcome measure of the self-reported stuttering severity there was no statistically significant

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[30] Assessment with GRADE.
Risk of bias: moderate (-1);
Indirectness: no evidence to warrant a reduction;
Imprecision: reduce by -1 as the size of the effect was highly uncertain for the outcome measures UTBAS and sociale phobia;
Inconsistency: n.a.;
Publication bias: no evidence to warrant a reduction.
[31] Stuttering percentage was determined by an independent speech & language therapist using two telephone conversations taped on a recorder. The first conversation was a routine discussion lasting 10 minutes. The second was a "surprise" conversation with a research assistant.

result for any of the variables. For the self-reported avoidance behaviour outcome measure there were two statistically significant results after 6 months: those with an anxiety disorder demonstrated more avoidance behaviour according to their own account than those with no anxiety disorder; the same applied for those with more than three psychiatric disorders.[32]

7.3.3 Studies into the Effectiveness of Cognitive and Behavioural Interventions Applied in the Netherlands in Recent Years

In 2004 Huinck and Peters published the results of a longitudinal (explorative) study of three stuttering therapies: the "Comprehensive Stuttering Programme (CSP)", the "Doetinchemse Method (DM)", and the "Individual Stutter Therapy (IS)". According to the researchers "the CSP particularly focuses on influencing speech behaviour via learning new speech techniques (controlled breathing, gradual glottal onset and delayed articulation). In addition, attention is also paid to the emotional and cognitive aspects of stuttering. In contrast to this, DM focuses primarily on the experiential aspects of stuttering and learning a more relaxed way of speaking with breathing-regulating exercises. DM, therefore, pays more time to emotions and cognitions whilst the CSP focuses more on "changing speech behaviour". Both therapies are offered as group therapies where the duration of and the protocol for the therapy are fixed. In contrast to the previous therapies, IS is individually focused where the "therapy programme is dependent on the nature of the individual stuttering problems and therefore the emphasis may be placed more on change in speech behaviour or the experience of stuttering without there being any time constraints".

Table 5 shows a number of characteristics of the therapy and the study participants. Two aspects are notable here. The number of participants in the various therapies is limited. Furthermore, there are considerable differences between the study participants in proportion to the duration of therapy, which may have had an effect on the outcomes in the Huinck & Peters study. The assessment time-points were chosen in order to evaluate the effectiveness of the therapies: immediately after therapy, after 1 year follow-up and 2 year follow-up. The two-year follow-up assessment is a strong point for this study. Insofar as where this can be assessed, standardised instruments were used to assess the outcome measures (stuttering percentage and speech rate; speech quality; speech motor system; emotional, social and cognitive factors associated with speech). Based on the second follow-up assessment the percentage of dropouts was 3.8, 6.7 and 48 for CSM, DM and IS respectively. The large

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[32]The degree of certainty around the effect size is limited. The statistical analysis was below far for the study. Therefore the risk of bias has been decreased by -2.

percentage of dropouts, particularly in the IS group, undermines the reliability of the study results.

Table 5. A number of characteristics of the therapy and the study participants according to Huinck & Peters (2004)

	CSP	DM	IS
Number of study participants	26	15	25*
Number of contact therapy hours per person	25.2	28.6	20.8
Percentage time focused on speech behaviour	73.3	36.1	32.9
Percentage time focused on emotional/cognitive aspects	26.7	64.9	67.1
Percentage breakdown by educational level:			
-lower and middle professional education	32	80	60
-higher general secondary education	12	7	0
-degree	56	13	40
* 10 participants dropped out in the therapy phase.			

Table 6. Various outcomes for the three different stuttering therapies according to Huinck & Peters (2004)

	CSP	DM	IS
Baseline stuttering percentage (1)	13.2	8.6	10.2
Stuttering percentage after 1 year (2)	8.0	6.2	6.1
Stuttering percentage after 2 years (3)	6.9	6.1	6.1
*Percentage improvement of stuttering percentage (3 w.r.t. 1)**	***91***	***41***	***67***
Baseline SPM (1)	126	138.2	135.0

	CSP	DM	IS
SPM after 1 year (2)	147.8	153.3	158.6
SPM after 2 years (2)	154.0	150.0	162.0
*Percentage improvement of SPM (3 w.r.t. 1)**	*18*	*8*	*17*
Baseline self-evaluation (1)	4.79	5.18	4.88
Self-evaluation after 1 year (2)	6.39	7.18	6.50
Self-evaluation after 2 years (2)	6.51	7.18	5.92
*Percentage improvement of self-evaluation (3 w.r.t. 1)**	*26*	*28*	*18*
Speech motor system - sequence /p t k/ DDK task			
Baseline	13.19	14.92	13.58
After 1 year	16.75	16.79	15.00
After 2 years	15.77	16.46	15.71
*Percentage improvement (3 w.r.t. 1)**	*16*	*9*	*14*

* Calculated by HdB

Table 6 shows the outcome measures of the different stuttering therapies. Care is warranted in the interpretation due to the limited numbers of study participants. Once again it looks like the CSP is the most effective of the three therapies. The "Individual Stuttering Therapy" appears to be more effective than the "Doetinchemse Method" for the majority of outcome measures (Table 6). Self-experience was an important outcome measure; this was a term used by Huinck & Peters (2004) to indicate the social, emotional and cognitive factors associated with speech. Self-experience was investigated using the following instruments:

- Perceptions of stuttering inventory (struggle, avoidance, expectancy)
- Brutten's speech situation list (emotional reaction, disordered speech)
- Erikson's communication questionnaire (S44, S24)
- Lanyon's stuttering severity questionnaire

- Inventory list concerning dealing with others - Tension (giving criticism, demanding attention, discussing your values, taking the initiative in conversation, valuing yourself and "total")
- Inventory list concerning dealing with others - Frequency (giving criticism, demanding attention, discussing your values, taking the initiative in conversation, valuing yourself and "total")
- Performance motivation test (performance motive, negative failure anxiety, positive failure anxiety)

Huinck & Peters (2004) represented the self-experience outcome measures as *absolute* differences between baseline and subsequent assessment (including follow-up). Given that the differences in the baseline assessments between stuttering therapies could (and would) lead to different outcomes, it would have been better to have included percentage improvement in order to determine the relative effectiveness of stuttering therapies. If we start by looking at the degree of statistical difference between the second follow-up assessment and the baseline assessment of the 23 assessed differences, 16, 9 and 7 are statistically significant for CSP, DM and IS respectively. The CSP also appears to be more effective here in comparison to the other stuttering therapies. It should, however, be noted that the CSP group had the most participants, meaning statistical significance was *in any case* more easily achievable.

Huinck & Peters (2004) provided a graphical representation for the results of the speech quality outcome measures (phonation, articulation, pitch, loudness and naturalness of speech). The scale used means that it is impossible to graphically interpolate the results. The degree of statistical significance has been provided in Table 7. CSP appears to be less effective here than either of the other therapies.

Table 7. Effects of three stuttering therapies on speech quality according to Huinck & Peters (2004)

	Statistical significance of the difference between baseline and the last follow-up assessment in proportion to stuttering therapy		
	CSP	DM	IS
Voice Dynamic factor	n.s.*	P=0.001	P=0.001
Articulation Quality factor	P<0.05	n.s.	n.s.
Speech Power factor	n.s.	P=0.001	P=0.001

The degree of certainty in terms of the effect size investigated in the Huinck & Peters (2004) study is limited, particularly due to the small numbers of study participants, meaning the outcomes are not very accurate, which entails that they are difficult to evaluate in terms of the presence/absence of clinical relevance. The second limitation referred to previously is the high percentage of dropouts in the IS group.[33]

Huinck's (2006) thesis explores the question whether the degree of stuttering severity and the severity of negative emotions and cognitions are associated with the diverse outcomes. Her conclusions are that there is no relationship between negative emotions and cognitions and that those who stuttered the most also enjoyed the most benefits from therapy, although they equally demonstrated the greatest degree of recurrence over the course of two years. Furthermore, she is of the opinion that the initial differences between the sub-groups with mild and severe negative emotions disappeared after treatment, "primarily as a result of a significant reduction in negative emotions and cognitions in the sub-group with severe negative emotions/cognitions". Huinck (2006) only conducted this analysis on the CSP group, and therefore we do not know whether the results obtained apply for the two other therapies as well.

Conclusion

Certainty of the effect size:	Various stuttering therapies, ranging from breathing therapy, "response contingent reinforcement" which incorporates "fluency shaping", the "Comprehensive Stuttering Programme'' (CSP), the "Doetinchemse Method" (DM), to the "Individual Stutter Therapy" (IS), have positive effects immediately after treatment on stuttering frequency, although the magnitude of the effect varies greatly. Where there was follow-up information available for at least six months after the end of therapy, some studies showed that the positive effects were maintained, whilst others demonstrated (some) recurrence.
limited for all relevant outcome measures	

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[33] *Risk of bias*: moderate (-1);
Indirectness: no evidence to warrant a reduction;
Imprecision: reduced by -1 as the size of the effect was highly uncertain;
Inconsistency: n.a.;
Publication bias: no evidence to warrant a reduction.

Those studies investigating Delayed Auditory Feedback (Lincoln et al., 2010, Ingham & Andrews, 1973) did not show any statistically significant difference in the percentage of stuttering between the experimental and control groups, nor was any follow-up undertaken.

There is insufficient evidence to apply the Alexander technique in the context of stuttering therapy. Adding videos of self-modelling or self-imposed time-out as therapies appear less effective than therapy based on restructuring speech.

The addition of cognitive behavioural therapy to therapy aimed at restructuring speech does not appear to have any added value to reducing stuttering frequency, however it does lead to a reduction in anxiety and avoidance behaviour and in problematic thoughts and beliefs about stuttering.

Both the telehealth and the standard version of the Camperdown Program provides around 60% in stuttering percentage after 12 months follow-up and around 40% in self-reported stuttering severity. Both of these programmes are equally effective. However the Camperdown programme has not been compared to other stuttering therapies in an RCT.

The certainty around the effect size is limited for the stuttering frequency outcome measure. This may partly be explained by the fact that it often concerns studies with few participants, meaning less accurate results are obtained. The great variation in types of therapies and study designs plays a role here as well. This has meant that the individual results of studies are difficult to combine, meaning that more accurate estimates of the effect cannot be obtained.

	Outcome measures other than stuttering frequency, such as avoidance behaviour (avoiding situations and words), naturalness of speech, participation or quality of life have often not been investigated and reported on in sufficient detail in order to make a judgment on the degree of certainty of the effect size.

Little research has been carried out to-date into variables (co-morbidity, socio-economics status, educational level, etc.) that influence the effectiveness of the interventions meaning that "patient-treatment matching" is still wishful thinking.

Studies into stuttering therapies applied in the Netherlands and evaluated by Huinck & Peters (2004) do not allow any firm conclusions to be drawn about the *relative* effectiveness although ***it appears*** that the "Comprehensive Stuttering Programme" is more effective than the "Doetinchemse Method" and "Individual Stutter Therapy" in terms of improving the stuttering frequency and better self-experience, although it is less effective in terms of improving speech quality (phonation, articulation, pitch, loudness and naturalness of speech). Taken as a whole, the "Individual Stutter Therapy" appears more effective than the "Doetinchemse Method".

No single treatment can guarantee that normal, fluent speech will be reached in the long term. |

Summary

The Herder et al. (2006) meta-analysis in particular demonstrated that breathing therapy and "response contingent reinforcement" are effective immediately after treatment for the outcome measure percentage of stuttered words or syllables. An effect size of 0.91 has been reported in comparison to no treatment, which significantly exceeds the 0.5 threshold required for a relevant effect. It should be noted here though that the effect sizes of the separate studies combined in the meta-analysis vary from 0 to 1.87. According to Herder et al. (2006) there appears to be no (large) differences in effect size for the various stuttering therapies based on the four included studies.

An extensive literature review by Bothe et al. (2006) revealed therapies that teach "prolonged" speech by effective forms of treatment. Bothe et al. (2006) did not report any effect sizes, but simply reported whether or not a therapy was successful in reducing the percentage of stuttered syllables or words to below 5% (an otherwise arbitrary threshold). If we limit ourselves to those studies reported by Bothe et al. (2006) providing follow-up data and including a comparative study design, then the reduction in stuttering frequency varies from 48 to 81% in therapies based on learning "prolonged" speech.

One study was included in the review by Woodman & Moore (2012) evaluating the effectiveness of the Alexander technique for stuttering frequency according to physical and psychosocial outcomes. This technique was designated by the reviewers as an alternative treatment method. The varying results of this study do not allow a definitive conclusion to be drawn. The telehealth version of the Camperdown programme was evaluated in a single study. This is a programme for restructuring speech with a focus on breathing, phonation and articulation. The outcome of this study showed that the telehealth version was potentially equally as effective as the face-to-face application of this programme (Carey et al., 2010). The addition of cognitive behavioural therapy to therapy aimed at restructuring speech does lead to a reduction in anxiety and avoidance behaviour and in problematic thoughts and beliefs about stuttering, but has no influence on the stuttering severity (Menzies et al., 2008). Video self-modelling, described by the researchers as "a behavioural intervention during which people view video images of themselves free of a problem target behaviour" does not appear, as a supplement to therapy based on restructuring speech, to reduce stuttering severity. Video self-modelling does perhaps have a beneficial effect on the quality of life (Cream et al., 2010). Self-imposed time-out (learning to just pause after a moment of stuttering) appears, as a treatment, to be less effective than therapy based on restructuring speech (Hewat et al., 2006). Those studies that investigated Delayed Auditory Feedback (Lincoln et al., 2010, Ingham & Andrews, 1973) did not show any statistically significant difference in the percentage of stuttering between the experimental and control groups.

In 2004 Huinck and Peters published the results of a longitudinal (explorative) study of three stuttering therapies applied by Dutch fluency specialists: the "Comprehensive Stuttering Programme (CSP)", the "Doetinchemse Method (DM)", and "Individual Stutter Therapy (IS)".

Due to the limited number of participants in the study, caution is warranted in the interpretation of the outcomes of the Huinck and Peters (2004) study. Furthermore this was

not a direct comparative study. Nevertheless, the CSP appears to be the most effective in terms of the reduction in stuttering frequency and improving emotional, social and cognitive factors as measured by standardised, self-completed questionnaires, followed by "Individual Stutter Therapy" (IS) and then the "Doetinchemse Method (DM)". However, CSP appears to be less effective than the other two methods in terms of speech quality (phonation, articulation, pitch, loudness and naturalness of speech). There do not appear to be any other relevant differences in respect to the other outcome measures (speech tempo, satisfaction with one's own speech, speech motor skills).

The literature review revealed that there were insufficient studies in terms of the avoidance behaviour (situation avoidance and word avoidance), naturalness of speech and participation or quality of life outcomes in order to determine how effective the various stuttering therapies are. In terms of the extent to which the effects of the various stuttering therapies on stuttering behaviour, participation and quality of life are maintained in the long term (at least 2 years) this cannot or can barely be quantified on the basis of the uncovered studies. There appears to be some recurrence in those studies in which follow-up details for 6 to 12 months after treatment were reported.

Information in respect to the question to what extent age, gender and co-morbidity influence effectiveness is virtually absent. There is evidence to suggest that the concurrent presence of multiple psychiatric disorders (anxiety disorders, personality disorders, mood disorders) may perhaps limit the effectiveness of stuttering therapy on one or more outcome measures.

The degree of certainty in terms of the effect size of the interventions is low for the majority of outcome measures. This can - to a large extent - be explained by the fact that it often concerns studies with few participants, meaning less accurate results are obtained. There is also a large variation in the types of therapies and study designs, meaning the individual results of the studies are difficult to combine.

7.4 From Evidence to Recommendations

7.4.1 Certainty in Respect to the Effect Size

In general it may be concluded that the effect size is uncertain for the majority of outcome measures of the stuttering therapies discussed above. This is due to the number of studies

(RCTs) being limited and the studies in terms of numbers are small. From this perspective there is no reason to prefer one of the stuttering therapies discussed above the others.

7.4.2 Balance of Desired and Undesired Effects

From the literature review, the frequency of stuttering (stuttering percentage) appeared to be the most reported outcome measure. Outcome measures other than stuttering frequency, such as avoidance behaviour (situation and word avoidance), naturalness of speech, participation or quality of life appear not to have been sufficiently investigated as a result.

In daily practice, adults and adolescents who stutter follow different types of help for their stuttering, for instance paramedic care or unconventional, commercial stutter programmes. Paramedic care is provided by a speech & language therapist or fluency specialist. This "tailor-made" care is provided on the basis of a treatment plan created in agreement between the speech & language therapist/fluency specialist and the PWS, and is appropriate to the help requested and the presenting problem. The speech & language therapist/fluency specialist will satisfy the requirements of the Paramedic Quality Registry. Non-standard, commercial stuttering programmes are often based on a specific view or approach to stuttering, often (but not always) based on personal experience with and solutions to stuttering. The programme is, broadly speaking, the same for all participants. Leaders or coaches of non-standard stutter programmes do not have to satisfy any quality standards. Moreover, no data as to the real effect of these various modes of stutter programs have been published in scientific litterature.

The desired effect of care provided by a speech & language therapist is for the PWS to receive custom-made care. Undesired results may include a lack of training on the part of the speech & language therapist in terms of treatment options, he/she may not recognise his/her own limitations resulting in the PWS investing too much time and energy in a particular approach and being referred on too late.

The added value of help offered by those who have experienced stuttering is the shared experience of being a (former) sufferer. An undesired effect is that what may have worked for one PWS may not work for another; the former person who stutters may not be sufficiently trained in terms of diagnosis and treatment and may not recognise their own limitations. This may also result in the PWS being referred on too late.

Therapy focused exclusively on speech change does not lead to a reduction in anxiety or avoidance behaviour and does not ensure an increase in psychosocial functioning. Nor does any of this take place in the period after therapy (Menzies, 2008; Yaruss, Quesal, Reeves, et al., 2002). Anxiety and worry often persist after an approach such as this (Cream, Onslow, Packman & Llewellyn, 2003; Plexico et al., 2009). This is an undesired effect.

Undesired effects may also occur in the clinical setting if the therapy is either not tailored at all or not tailored properly to the PWS. In cases such as this the PWS often has difficulty using

(or continuing to use) the lessons learned in practice. Recurrence often takes place (Manning 2010, page 475-476, 530, 575-588).

This is different for cognitive behaviour therapy as this contributes to a reduction in anxiety, avoidance behaviour and problematical thoughts and beliefs about stuttering (Menzies, 2008). Cognitive behavioural therapy ensures for an improvement in the quality of life. This is a desired effect. The PWS does not (any longer) view stuttering as a problem, and therefore does not (any longer) have a social-emotional handicap. The PWS is fully able to participate in society and no longer avoids activities because of their stuttering. This is a common experience in practice and the clinical setting.

The desired effect is for the tailored treatment aims adapted to the specific problems and the help requested by the PWS are achieved.

Both PWS, as well as speech therapists/fluency specialists who are familiar with participation in group therapy acknowledge that the activities and opportunities of group therapy can be an appropriate supplement to individual treatment. To-date not much research has been undertaken into the effects of group therapy. However, a number of studies have suggested that group therapy (Stewart & Richardson, 2003; Hearne et al., 2008; Beilby et al., 2013) and self-help groups (Boyle, 2013) make a positive contribution to treatment results. The Huinck and Peters (2004) study suggests a certain degree of relative effectiveness in terms of group therapy. The collective setting enhances the learning and growth effect in terms of promoting and maintaining learned skills and insights (Manning, 2010, page 411). Turnbull (2005) also reports the strength of participating in group therapy: "It is our experience that many clients who participate in group therapy record more progress and better results in terms of achieving and maintaining behaviours than clients in a one-on-one setting". (Turnbull & Stewart translated 2005, page 136) (Stewart, 1996). Children appear to prefer group therapy when they are looking for help (Hearne, 2008) and a combination of individual and group therapy provides a significant effect for young adolescents (Fry, 2009).

Scientific research has shown that the use of equipment based on Delayed Auditory Feedback (DAF) does not ensure for a significant difference in the percentage of stuttering. In practice it was apparent that DAF has a transient effect for some PWS. However this is usually negated. DAF may have an added value for only some PWS. However it is important that the user realises that because the effect of DAF is transient it can only be employed sporadically. It is preferable for this to be undertaken in discussion with the speech & language therapist/fluency specialist.

As yet there do not appear to be any indications that applying the Alexander technique, video self-monitoring or self-imposed time-out have any added value.

7.4.3 Values and Preferences

Given on the basis of the RCTS published in the scientific literature that no single aid for stuttering has been *proven* to be more effective than any other, more should be done to satisfy the wishes and needs of people who stutter (PWS). These form the basis for the treatment plan drawn up on the basis of informed consent.

The working group is of the opinion the problems associated with stuttering extend beyond the percentage of stuttered syllables, as is often reported in a lot of scientific research. Given the variety of variables influencing the long-term effect of help and the unique combination of variables per PWS a multi-dimensional approach is the preferred treatment plan. Many authors who have written about stuttering therapy have found a multi-dimensional approach useful in the treatment of PWS (incl. Guitar, 2014; Manning, 2010 (p588); Yaruss, 2010). This is supported by the American Speech-Language-Hearing Association (ASHA, 2007). Quality of life also deserves attention alongside tackling core stuttering behaviours (including non-audible stuttering). Craig, Blumgart & Tran (2009) demonstrated that the quality of life for PWS is lower than in a control group of people who did not stutter. Applying the ICF model means that the stuttering problems can be tackled using a multi-dimensional approach; all of the elements together forming the stutter problem need to be treated.[34]

The ICF for speech/language pathology was developed in 2009. The Dutch Association for Logopedics and Phoniatrics (NVLF, www.nvlf.nl) has trained professionals in applying the ICF within treatment. The NVLF uses the ICF body of thought in developing guidelines and other quality instruments, such as speech & language standards, the professional profile and quality tests.

The working group is of the opinion that the speech & language therapist/fluency specialist should make use of all the ICF elements with the PWS. Subsequently the PWS should be thoroughly informed about the various treatment options in terms of all of the relevant elements. The benefits and disadvantages should be clear and aimed at the PWS's specific circumstances. Ultimately the therapist and PWS should be able to agree on the basis of "shared decision making" which path to follow. This should be justified on the foundation of a treatment plan with SMART aims. Linking these aims to the ICF components produces the required opportunity to treat both the entire stuttering problem and evaluate the treatment.

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34 The *World Health Organization (WHO)* has developed an international classification system to categorise individual functioning. This is the *"International Classification of Functioning, Disability and Health" (ICF)*.

The working group is of the opinion that a speech & language therapist and/or fluency specialist should apply the body of work from the ICF and where possible work along evidence-based practice in his daily work.

- **Persons who stutter**

As there is an absence of significant or moderate certainty in respect to the effect size of various stuttering therapies for the majority of outcome measures, PWS preferences may play an important role.

A therapy with a good outcome comprises more than a reduction in the stuttering frequency and/or stuttering intensity. A therapy may be referred to as being successful if the outcome includes a reduction in the stuttering frequency and/or intensity, the PWS is satisfied with their speech and/or the PWS is confident to stutter freely. There is no easy "cure" for stuttering. The treatment needs to be tailored to each PWS. Therapy will cost the PWS a great deal of effort and take up a long period of time. The PWS may also stop and restart therapy at a later stage. Although the speech & language therapist/fluency specialist will link in to this process and help find the right path, the person who stutters will, however, have to do this him or herself. The speech & language therapist/fluency specialist will need to possess a broad range of skills in order to be able to adapt the treatment to the needs of the PWS. Furthermore it is also desirable that the speech & language therapist/fluency specialist utilises group therapy in addition to individual therapy (with or without the aid of another speech & language therapist/fluency specialist), so that the maximum help is available for the PWS. It is apparent in practice that many people who stutter value and learn from having contact with others who stutter. This is evident from a focus group, which is the experience of Demosthenes members and is underlined by the Boyle (2013) study.

A therapy should contain more than learning a speech technique and/or other physical exercises. There should also be work on the perception of and feelings arising with the stutter. This is the most important outcome of the focus group in terms of the contents of the therapy and is also apparent from research with PWS (Yaruss, Quesal & Murphy, 2002). Usually, it is only possible to deal well with stuttering in daily life, if the person who stutters is able to think and talk rationally about it. Dealing with stuttering in daily life (and not just in the therapy setting) has to be an essential part of therapy.

Satisfaction with speech is usually only be achieved by the PWS taking responsibility themselves for their stuttering, getting out the house and not letting their life be limited.

- **Speech & language therapists - Fluency specialists**

Speech & language therapists in the Netherlands are trained to treat patients with fluency disorders. However, not all speech & language therapists feel competent enough in terms of treating stuttering. Speech & language therapists who are members of the Dutch Association for Stutter Therapy (NVST) have specialised in stuttering; they work, where possible, along

evidence-based lines and usually have more knowledge, skills and experience in the field of treating fluency disorders than a general speech & language therapist. They have been trained both in working on the technical aspects of speech, as well as on the psychological, pedagogical and psycho-social problems that are regularly associated with a patient who stutters. They follow the international consensus of using an integrated approach and are expected, as part of their specialisation, to be trained to provide group therapy as an addition to individual therapy.

Currently, different forms of group therapy are provided in the Netherlands by speech & language therapists/fluency specialists. There are forms of intensive group therapy (Bezemer et al., 2006; Weijts et al., 2012) but in addition to this other forms of group therapy are also provided in clinical practice, such as exercise groups, parent groups and children groups.

There is no evidence that the stuttering therapies, as provided in the Netherlands by speech & language therapists/fluency specialists, needs to be modified. Furthermore, there does not appear to be a demand to become competent in a different or a specific form of stuttering therapy. There needs to be "shared decision-making" in the choice of therapy, and that the person who stutters should be referred to as required. This also means that the practising speech & language therapist/fluency specialist is aware that there are three overarching factors in all clinical procedures that contribute to a successful therapeutic outcome: the "working alliance" between the PWS and the therapist, the degree of flexibility the therapist attaches to and implements a treatment protocol and the quality (level of expertise) of the clinician (good therapeutic skills) (Ahn & Wampold ,2001 in Manning (2010, p 310)).

Training fluency therapists in cognitive behavioural therapy (CBT) has been shown to be useful. Although the evidence for this is not strong, CBT may be of significance, particularly for people who stutter wanting to reduce feelings of anxiety and avoidance behaviour. Recent research reports that Acceptance and Commitment Therapy provided in a group setting could well be a new form of intervention. The training of speech & language therapists/fluency specialists in group settings as a supplement to the one-on-one setting is also desirable as both (early) research and practice have shown that this provides added value.

The specialisation "fluency specialist" has a clear place in speech & language therapy in the Netherlands as a follow-up to the initial training of a speech & language therapist and emphatically contributes to the education of the "expert" speech & language therapist/fluency specialist.

7.4.4 Costs of Therapy

There are no exact figures known about the costs of treating stuttering, partly due to the variety of the therapeutic techniques applied. It is difficult to put a figure on the extent to which the following recommendations would have an effect on the costs associated with stuttering therapy, even more so as there is no clear preference that can be provided in respect to certain stuttering therapies or techniques.

Therefore the working group has been unable, when drawing up the recommendations, to put an exact figure in terms of the costs. Further research into the cost-effectiveness of stuttering therapy is desirable given the developments in healthcare.

Recommendations

13. The treatment of stuttering for adolescents and adults should be individualized. Therapy may take place in an individual and/or group setting. The treatment plan is established in a dialogue between the SLT and the Person Who Stutters (PWS) (' shared decision making '); the treatment plan contains all ICF-elements. The wishes and needs of the PWS form the basis of that treatment plan.
14. Depending on what has been agreed by the person who stutters and the speech & language therapist/fluency specialist, the treatment will focus on the psychosocial aspects (emotional; and cognitive reactions to speaking), on verbal-motor aspects or on both. Cognitive behavioural therapy is recommended for the treatment of psychosocial aspects.
15. Other elements of stuttering therapy should include: *Promoting transfer* - it is essential that the PWS applies the skills learned and insights in daily living. *Promoting and maintaining self-management* - the PWS is able to evaluate the stuttering and the associated behaviour and adjust this if necessary.
16. In general, the use of Altered Auditory Feedback based devices is not recommended. However, in specific circumstances, such equipment may reduce the stuttering rate in some PWS. One should note that this effect may not be maintained.

Rationale for the recommendations
- Various stuttering therapies have positive effects directly after treatment on the stuttering frequency; these, however, vary in impact. Some studies report that positive effects are maintained six months after therapy, yet others report (some) recurrence. The certainty around the effect size is limited for the stuttering frequency outcome measure, through limitations in study design and sample size. Outcome measures other than stuttering frequency, such as avoidance behaviour (situation and word avoidance), naturalness of speech, participation or quality of life have not been sufficiently investigated.
- The scientific literature does not provide any strong evidence that one stuttering therapy is (much) more effective than another.

- It is therefore appropriate to jointly determine the choice of a specific therapy based on the wishes and needs of the person who stutters.
- The working group, in drafting these recommendations, has been unable to attach any conclusive figures on the costs of stuttering therapy, due to the lack of information about the current costs.
- The recommendation for therapy in a group setting rests on the experience of members of the working group and the experiences of persons who stutter, as expressed in the context of a focus group organised as part of this guideline. Therapy in a group setting may enhance the motivation for therapy and be a supplement to peer-group contact.

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Chapter 8: Effectiveness of Pharmacotherapy for Adolescents and Adults

8.1 Introduction

Neither the cause of stuttering nor the cause of individual moments of stuttering have been established with any certainty (see also the introduction of this guideline). There have been numerous attempts to identify effective pharmaceuticals. This chapter discusses the current state of affairs. There will only be a brief description of the current state of affairs given that pharmacotherapeutics are not applied in the Netherlands. Any evaluation in respect to the certainty of the effect size is therefore also absent.

8.2 Method

A search was undertaken for systematic reviews/meta-analyses and individual RCTs where pharmaceuticals were evaluated in terms of effectiveness and safety. Please refer to paragraph 2 of the text describing non-pharmacological interventions for a description of the search strategy. Two relevant reviews and two relevant RCTs were found.

8.3 Evidence

We will firstly discuss the systematic reviews by Bothe (2006) and Boyd (2011). After which the individual studies that have been subsequently published will be discussed.

8.3.1 Overview of the Systematic Reviews

Bothe et al. (2006)

In 2006 Bothe et al. published a systematic review of studies investigating the effectiveness and safety of pharmaceuticals. These pharmaceuticals comprised:

- anticonvulsives: carbamazepine
- antidepressants: phenelzine, paroxetine, sertraline, mianserin, clomipramine and desipramine
- antipsychotics: haloperidol, olanzapine, risperidone
- cardiovascular medicines: clonidine, oxprenolol, propranolol, verapamil

- cholinergics: bethanechol
- dopamine antagonists: pimozid, tiapride
- neuromuscular blockers: botulin toxin
- anxiolytics + antidepressants: benzodiazepine with citalopram

Of the 35 studies:

- there was one (risperidone) which demonstrated a reduction in the stuttering frequency to below 5%; information about stuttering frequency prior to the administration of risperidone was not reported by the authors;
- there were three (haliperidol, propranolol, sertraline) which indicated a reduction of more than 50% in stuttering frequency;
- there were four (botulin toxin, clomipramine, desipramine, carbamazepine) which demonstrated an improvement in the cognitive, social or emotional domains.

Given these results the effectiveness of pharmacotherapy appears limited. It should be noted that pharmacotherapy is often associated with side-effects. These side-effects may be mild, such as a dry mouth, constipation, fatigue or weight increase. They may also be severe, such as Parkinson-like symptoms resulting from haloperidol or seizures caused by tricyclic antidepressants.

Boyd et al. (2011)

Boyd et al. (2011) conducted a review where they included studies that specifically pertained to the age group younger than 18. They found seven studies, of which two were case studies, which they categorised as very weak evidence. They characterised four studies as weak evidence because two essential quality criteria had not been satisfied: the presence of a control or placebo group, and data about the dysfluency prior to and after the intervention. One study (Althaus et al., 1995) was described as strong evidence since it was a double-blind, placebo-controlled cross-over trial. The age of participants in this study (N=25) varied between 6-13 years. Of these 25 children six also had ADHD, whilst four other children also had a developmental disorder or a chronic motor disorder.

The medicine that was evaluated was clonidine which was taken for eight weeks in a quantity of 4 µg/kg body weight per day. Dysfluencies were measured in terms of the number of repetitions, prolongations, blocks and interjections. The children were asked to read a text out loud, repeat sentences read out loud, and to say something about their experiences during a weekend or holiday. The precise measurement is not apparent from this study, nor is it clear which units were used for the outcomes. For those receiving clonidine, the frequency of repetitions and extensions increased: the difference between follow-up and baseline assessment was 2.33 (95% CI: -4.7; 9.3) and 0.58 (-0.22; 0.79) respectively. The width of the confidence intervals also demonstrated that the results obtained were not very accurate. A number of other effects resulting from the clonidine administration were, however, significant

and of clinical interest according to the authors: on the basis of a behavioural checklist parents and teachers reported that the children were less hyperactive, more task-focused and more easily approachable.

8.3.2 Individual Studies into the Effectiveness and Safety of Pharmaceuticals

Maguire et al. reported a study in 2010 concerning the effectiveness and safety of

Pagoclone.[35] (NB HdB: For the text below use was made of the review by Dr Bert Bast, see http://www.stotteren.nl/professionals/wetenschappelijk-onderzoek/alge-meen/136-stotterpil.html).

This study was sponsored by the pharmaceutical industry. The primary outcome measures were the usual objective and subjective "stutter severity scales" (SSI-3, SSS, SEV, stuttering percentage). The secondary outcome measures were naturalness of speech and the Liebowitz Social Anxiety Scale. Following the baseline assessment there were another 2 or 3 assessments: after 2, 4 and 8 weeks from the start of treatment. In virtually all cases outcomes improved in both the placebo group and the Pagoclone group. In only a few cases was there a significant difference in favour of Pagoclone. The strongest impact was measured in the percentage of stuttered syllables (stuttering percentage); this appeared to have been reduced by 21% after 4 weeks of Pagoclone (see Table 1 for effect sizes). The placebo-controlled, double-blind study was followed by a so-called open-label study. The improvement in the stuttering frequency (percentage of stuttering) was apparent after 1 year in 40% of the Pagoclone group. However, if these relative percentages are converted into absolute percentages, there is an average reduction in the percentage of stuttering from 8.3% to 6.7%. This appears to barely be of clinical significance, particularly when the side-effects are taken into account (fatigue, headache).

Busan et al. (2009) investigated the effectiveness of paroxetine in terms of the reduction in stuttering frequency and movements associated with stuttering. Five persons who stuttered received 20 mg paroxetine once per day during 12 weeks, and another 5 persons who stuttered received a placebo. After 12 weeks the stuttering frequency in the paroxetine group decreased in absolute terms by 5.7% (median) and in relative terms by 45.2%; the corresponding figures in the placebo group were 1.6% and 15.8%. The percentage of stuttering was determined on the basis of spontaneous speech and reading a passage of text. No statistically significant differences were determined between the two groups. Stutter-related movements (facial) - strictly speaking not a relevant outcome measure - did appear to

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[35] The pharmacist stopped with further research into the effectivenesss of pagoclone.
http://thestutteringbrain.blogspot.nl/2011/12/breaking-news-endo-stops-pagoclone.html.

be significantly less. The investigators did not focus on the occurrence of side-effects of paroxetine. According to the pharmacotherapeutic formulary (consulted on 4 June 2013) there are the following side-effects: in more than 10% of cases: nausea and sexual disorders; in 1-10% of cases: increase in cholesterol level, drowsiness, sweating, muscle weakness, insomnia, agitation, abnormal dreams, dizziness, sensory disorders, tremor, visual disorders, yawning, constipation, diarrhoea, dry mouth, asthenia, increased body weight, anxiety, headache and reduced appetite; in 0.1-1% of cases: abnormal bleeding (primarily skin and mucosa), confusion, hallucinations, extrapyramidal disorders, mydriasis, tachycardia, transient increase or reduction in blood pressure, postural hypotension, skin rash, itch, urine retention and urinary incontinence.

Table 1. Effectiveness of Pagoclone compared with a placebo

Outcome measures		Large effect size	95% CI
SSI-3 Frequency and Duration Subscore			
	Change to wk 4	0.52	0.13-0.90
	Change to wk 8	0.21	-0.17-0.58
SSS Severity Subscore			
	Change to wk 2	0.40	0.02-0.78
	Change to wk 4	0.20	-0.18-0.58
	Change to wk 8	0.14	-0.24-0.51
Site clinician-rated SEV			
	Change to wk 2	0.20	-0.18-0.58
	Change to wk 4	0.36	-0.02-0.74
	Change to wk 8	0.13	-0.25-0.50
Percentage of syllables stuttered			
	Percent change to wk 4	0.62	0.24-1.01
	Percent change to wk 8	0.28	-0.10-0.66
LSAS (Liebowitz Social Anxiety Scale Total)			
	Percent change to wk 4	0.22	-0.14-0.58
	Percent change to wk 8	0.18	-0.19-0.54
LSAS Fear/Anxiety Subscale			
	Percent change to wk 4	0.15	-0.22-0.51
	Percent change to wk 8	0.06	-0.31-0.42
LSAS Avoidance Subscale			
	Percent change to wk 4	0.26	-0.10-0.62
	Percent change to wk 8	0.25	-0.11-0.61

LSAS Stuttering Subscale			
	Percent change to wk 4	0.30	-0.07-0.66
	Percent change to wk 8	0.23	-0.13-0.60

Source: Maguire et al. (2010)

Conclusion

Certainty in respect to the effect size: **Not ascertained**	The majority of studies into the effectiveness of pharmacotherapy have not demonstrated any clinically relevant effects on stuttering frequency and outcomes of a social cognitive or emotional nature. Given the potential side-effects of pharmacotherapy which may have a negative impact on quality of life, the net effect seems to be zero.

8.4 From Evidence to Recommendations

8.4.1 Certainty in respect to the Effect Size

Although the quality of evidence has not been ascertained, it may, however, be noted that many studies into the effect of various pharmaceuticals on stuttering are methodologically weak. It may also be noted that usually no statistically significant effect is seen.

8.4.2 Balance of Desired and Undesired Effects

Virtually all pharmaceuticals are associated with side-effects after long-term use. Given the absence of significant effects of pharmaceuticals on the one hand and the potential of side-effects on the other, there is no reason to recommend the long-term application of pharmaceuticals.

8.4.3 Values and Preferences

a. Persons who stutter

There is no literature about the effects of temporary use of medication. A call via Demosthenes regarding the effects of temporary use of medication did not deliver a response.

b. Speech & language therapists - fluency specialists

When pharmaceuticals have been used in stuttering where there is co-morbidity present, it is of clinical importance that the therapist is aware that behaviour may improve while stuttering severity increases or that stuttering may be evoked by the pharmaceuticals. The latter cases are referred to as acquired stuttering or pharmacogenic stuttering (see the Introduction).

Recommendation

17. Use of pharmaceuticals in the context of stuttering therapy is not recommended. Where there is co-morbidity and stuttering, it is recommended that an appropriate choice and dose of pharmaceuticals is sought in consultation with the PWS (and their direct environment) and the prescriber.

Rationale for the recommendations

- Virtually all pharmaceuticals are associated with side-effects after long-term use. Given the absence of significant effects of pharmaceuticals on the one hand and potential side-effects on the other, there is no reason to recommend the (long-term) use of pharmaceuticals.

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Chapter 9: When and for what Reasons should a Patient who Stutters be Referred by a Speech & Language Therapist to a Fluency Specialist or another Healthcare Provider?

9.1 Introduction

In many cases the request for treatment in association with stuttering is firstly raised with a speech & language therapist. However, the latter may not always feel he/she has the right degree of skills in respect to all of the aspects of this treatment. Therefore there are frequent referrals on to a fluency specialist. Occasionally there may be excessive delays, perhaps due to a lack of knowledge about the expertise and treatment options offered by a fluency specialist.

Fluency specialists are speech & language therapists specialising in stuttering, who after undergraduate training in speech & language therapy have completed additional postgraduate training approved by the NVST (Dutch Association of Stuttering Therapy). The differentiation between speech & language therapists and fluency specialist has come about through the need to improve the quality of care of the PWS and enhance their skills base. The effect of stuttering on social/emotional functioning and the quality of life of the PWS means treatment is complex. Cognitive and emotional training form a part of therapy, as does the mentoring of the family of a child who stutters. The basic training in speech & language therapy does not sufficiently provide for this.

The criteria for referral to the appropriate healthcare provider should be clear, both to the family doctor and the individual seeking help.

Severe speech anxiety or social anxiety may arise in PWS, sometimes this is associated with depression symptoms. Social anxiety may also be (or have become) the principal problem and stuttering an additional issue. In that case the help of another healthcare provider may be desirable. This chapter provides more clarity about when a referral should be made and to which healthcare provider.

9.2 Method

No systematic literature reviews have been undertaken in order to answer the principal question of this chapter. In terms of describing the patient perspective, use has been made of the report from a focus group meeting of PWS ((CBO), 2013). For the description of the

professional perspective, use has been made of the knowledge, experience and opinions of the working group members acting as speech & language therapists/fluency specialists.

9.3 Evidence

Stuttering is a complex disorder, particularly because social/emotional problems occur relatively frequently. Detailing this is therefore of importance. Treatment of stuttering does not always lead to sufficient improvement in the expected treatment period. Referral from a speech & language therapist to a fluency specialist, or referral to another healthcare provider may be useful due to the social/emotional problems and inadequate realisation of the treatment objectives. This is explored in further detail below.

9.3.1 Social/emotional Problems

Stuttering may, to a great extent, impact on communication and thereby also on the social and emotional well-being of the PWS. For older children and adults who stutter, the communication problems may become so great that they impact on social life, education and career choices. Some PWS develop serious emotional problems such as depression or social anxiety (Blumgart, Tran & Craig, 2010; Iverach & Rapee, 2013; Koedoot, Bouwmans, Franken & Stolk, 2011; Tran, Blumgart & Craig, 2011). During the diagnostic process in speech & language therapy the PWS will be asked about anxiety avoidance behaviours. Questionnaires will be completed detailing anxiety about speaking and speech situations; these include the Behaviour Assessment Battery (BAB) (see research question 2). If it is apparent that the anxiety experienced is not related to the stuttering or if the anxiety is beyond proportional then there is a possibility of social anxiety according to the diagnostic classification system for psychiatric disorders (DSM-5) (Iverach & Rapee, 2013).

It is recommended to ask the PWS or Child Who Stutters (CWS) about potential depression symptoms during the diagnostic phase or during therapy if there are indications to do so. In the Multidisciplinary Depression Guideline (2013) there is a discussion of the questionnaires used to detect depression. For children aged 3 to 13 the Strength and Difficulties Questionnaire (SDQ) is recommended. This is freely available (www.sdqinfo.org).

The Four Dimensional Symptom List is recommended (4DSL) for adolescents and adults. The Depression Recognition Scale (DRS) is an instrument that can also be used as an initial screening tool for depression symptoms (Ruiter & Jong, 2010).

Children and adolescents have a greater risk of being bullied and experiencing negative reactions to their stuttering from their peers. This has consequences for their self-worth and makes them vulnerable to developing psychological problems during adulthood (Blood et al., 2010; Langevin, 2009). The speech & language therapist/fluency specialist that has regular contact with the child or adolescent should be alert to bullying behaviour. This may be achieved by asking about bullying. The following questionnaire drawn from the draft Paediatric Healthcare Guideline (JGZ Guideline, 2012) may help as a guide here:

Questions to detect bullying:

Do you feel good? How do you feel at school?

Bullying:

Have you recently been pestered or are you sometimes not allowed to take part in the group?

Bullying:

Have you recently bullied someone, laughed at them or told them they were not

allowed to take part in the group?

Cyberbullying:

Do you sometimes receive something annoying via your mobile telephone, internet or online gaming?

Do you send negative messages to others?

Accomplice:

Do you sometimes take part in bullying other children?

Consequences of bullying:

Are you frightened or sad? Have you got a headache, stomach ache, little appetite? Do you sleep well?

Additional diagnostics and/or individual care is indicated where there are signs of potential bullying. The JGZ Bullying Guideline (2012) emphasises the importance of parental involvement in determining the care pathway and encouraging parents to discuss the bullying issue at school.

9.3.2 Course of Therapy

The speech & language therapist employs a methodical and step-wise approach to therapy. A part of this is regularly evaluating the stated treatment goals with the PWS and/or their direct environment, usually every two to three months.

A significant improvement in stuttering occurs in the treatment of young children up to the age of six who stutter on average after 11-12 treatment sessions (Kingston, Huber, Onslow, Jones & Packman, 2003; Millard, Nicholas & Cook, 2008; Yaruss, Coleman & Hammer, 2006). This pertains to treatment using the Lidcombe Program and the Demands and Capacities model or a similar type of model[36]. The effect of these interventions is large in comparison to non-treatment (refer to Chapter 3, research question 3c).

In older children and adults there is less clarity about the anticipated course of therapy over a given time period. There are many variables that play a role in the therapy process, such as the age of the PWS, the intensity and frequency of the therapy offered, the degree of secondary problems, such as anxiety and avoidance behaviour and the support from the PWS's environment (Manning, 2010).

An open dialogue is important during the joint evaluation of the therapy process and the interim result, in which the PWS is free to be able to express his/her views and thoughts without reservation. This will enable a decision to be taken along with the speech & language therapist/fluency specialist ("shared decision-making") about the steps to be taken in the therapy.

9.4 From Evidence to Recommendations

a. Persons who stutter

In the focus group, PWS and parents of children who stutter indicated that they often receive inadequate information about the treatment options in the Netherlands. They want to be informed about the existence of fluency specialists and about organisations such as the Dutch Stutter Federation, the stutter information centre (SIC) and the stotteren.nl website.

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[36] This pertains to the Parent-Child Interaction Therapy (PCIT) and the Family-focused Treatment Approach

Good information provision to parents and PWS is necessary in order to be able to consider and determine what care and what type of care provider are the most appropriate.

Treatment close to home is preferable for patients. Long travel journeys may be a significant burden for the PWS or for the family of a child who stutters. Furthermore, the benefit of having help close to home is that the care provided is aware of the social environment and probably has contacts with healthcare organisations and schools in the neighbourhood.

The focus group expected the speech & language therapist to show an understanding of the problems around stuttering in a broad context, including the social and emotional factors. Should a referral to another professional be necessary, then people would like to hear that on time and be supported in the choice of the right healthcare provide. People sometimes feel they have been left hanging around for too long.

This same point was made by the focus group about the GP and paediatricians' policy: too little understanding for the social/emotional factors of stuttering and referrals that are too late were highlighted numerous times as bottlenecks. One of the tasks of the speech & language therapist/fluency specialist is to inform GPs and paediatricians in their professional network about stuttering, the criteria for referrals and treatment options.

b. Speech & language therapists - fluency specialists

The most important differences between a speech & language therapist and fluency specialist are provided below (Bezemer, Bouwen & Winkelman, 2008):

The speech & language therapist:

- has basic knowledge about stuttering
- is able to provide a lot of information "relevant" to the client
- is usually notified at the first signs, when there is still little acquired secondary behaviour
- is often close to the child's world, probably knows the school or nursery
- has a lot of knowledge about the entirety of speech and language development
- is used to employing many developmental aspects in research/therapy
- has access in an increasing degree to recent information about stuttering via, amongst others, post-professional training courses and literature reviews

Fluency specialists:

- have current knowledge about the latest research data concerning stuttering
- have a rich range of therapy approaches for each developmental phase. Therefore a considered therapy choice may be made for each client

- are able to adapt the approach chosen to the client, working eclectically to produce the required tailored method
- have more children who stutter in therapy meaning other therapy formats are available such as: one hour sessions (in contrast to half hour sessions with the speech & language therapist), group therapy, parent guidance in groups, teacher guidance. This allows different forms of learning to take place
- possess a broad range of tools and services for research and treatment of stuttering and develops, where required, new strategies and materials
- are aware of the mutual relationship between stuttering and other disorders
- offer the option of a second opinion to the client

The differences between the speech & language therapist and fluency specialist are, of course, being deliberately magnified in this publication, and speech & language therapists may in some cases have the same characteristics as fluency specialists.

It is desirable when providing therapy to a PWS that the speech & language therapist or fluency specialist are able to implement the approaches in this guideline at a minimum. After all, this guideline has recommendations in respect to the optimal logopaedic care to people with a stutter in terms of diagnostics, treatment, referral and follow-up care taking due consideration of the current state of scientific literature and insights within professional practice in 2013. This means that psychosocial aspects such as (speech) anxiety, avoidance behaviour and verbal-motor aspects of stuttering are potential therapy goals. It may be expected from a speech & language therapist that they have insight into their own therapeutic capacity and approaches, and that they take professional responsibility to take measures if he/she knows or notes that his/her own expertise is falling short.

The speech & language therapist may make referrals to a fluency specialist, and may also request a (telephone) consultation or a coaching session in planning and implementing therapy.

Agreement and transfer during (re-)referral

When there is a referral from a speech & language therapist to a fluency specialist, or a referral to another healthcare provider due to social/emotional problems and an inadequate realisation of the treatment goals, this is undertaken with a clear request. The referring therapist will ask for the PWS's consent to inform the GP and colleague about the details from the case history, investigation, therapy and reason for referral.   It is also recommended that the data and advice from the various healthcare providers agree

with each other and the referring therapist stays within the competences or area of competence of the healthcare provider to whom the referral is being made.

The recommendations for the referral and collaboration as formulated in this guideline may be converted into working practises regarding referrals of PWS after local or regional agreement. The National First Line Collaborative Agreement (LESA) contains questions that may be of particular relevance for this (Faber et al., 2008). The working group has adopted these questions.

These questions may be used as a discussion point when drafting local or regional working practise agreements about referrals for stuttering.

- When should patients be referred to particular therapists?
- Does an interim evaluation of the treatment take place with a GP referral? What agreements have been put in place for this?
- How is mutual agreement achieved for the working practises and responsibilities?
- What agreements have been made about reporting and re-referrals?
- Does the information materials and advice from healthcare providers agree with each other?
- Which therapists (speech & language therapist, fluency specialist, other therapists) are there available in the region (Faber et al., 2008)?

Costs

It is not exactly clear what the costs are of a timely or delayed referral to an appropriate healthcare provider or therapist, however it is plausible that continuing with therapy without the desired outcome will increase costs.

Recommendations

Expertise

18. The speech & language therapist needs to be able to implement the recommendations in this guideline regarding diagnostic and therapeutic approaches. If the SLT lacks the experience or knowledge to do so, the client should be referred to an SLT with more expertise in the field of stuttering or to a Fluency Specialist.

Social/emotional problems

19. Where there is a suspicion of social anxiety or depression in the person who stutters based on findings from the Behaviour Assessment Battery (BAB), the speech & language therapist or fluency specialist needs to consolidate this with the Strength and Difficulties Questionnaire or the Four Dimensional Symptoms List. Following a positive indication the speech & language therapist or fluency specialist should consult with the PWS or the parents about a referral to the GP for a potential referral to a psychologist or psychiatrist.
20. In case of stuttering children and young people, the SLT should explore the problem of bullying. This may include a suitable questionnaire for bullying. If bullying is identified, the SLT should discuss this with the parents and agree a plan of action.

Course of therapy

21. During the assessment, the SLT will provide information to the PWS regarding the treatment options in order to enable an informed choice regarding the treatment. The SLT should provide information regarding other relevant resources and websites.
22. If a child who stutters up to aged 6 years has not made progress with 11 to 12 therapy sessions or within 3 months, a Fluency Specialist should be consulted.
23. If during two-or three-monthly evaluations of the therapy process, the realistic and achievable treatment goals may have not been sufficiently realized (according to the PWS or to the SLT) the SLT should discuss the possibility of referral to another practitioner.

24.	The SLT and other professionals involved with PWS in a specific region set up a working agreement concerning collaboration, referring and information transfer. When referring to another SLT or Fluency Specialist, the SLT formulates specific questions incorporating all relevant ICF elements.

Rationale for the recommendations

- The complexity of stuttering and the associated social/emotional problems requires specific skills from the therapist.
- Continuing with a therapy that is having an inadequate effect is undesirable.
- In the treatment of young children aged up to six who stutter a significant improvement in stuttering occurs on average after 11-12 treatment sessions (Kingston, Huber, Onslow, Jones & Packman, 2003; Millard, Nicholas & Cook, 2008; Yaruss, Coleman & Hammer, 2006).
- Some PWS develop serious emotional problems, such as depression or social anxiety (Blumgart, Tran & Craig, 2010; Iverach & Rapee, 2013; Koedoot, Bouwmans, Franken & Stolk, 2011; Tran, Blumgart & Craig, 2011).
- Children and adolescents who stutter have a greater chance of being bullied and of experiencing negative reactions from their peers (Blood et al., 2010; Langevin, 2009).
- The fluency specialist has additional expertise in treating stuttering.
- The speech & language therapist or fluency specialist are not equipped to deal with complex or severe social/emotional problems irrespective of whether this is caused by stuttering.
- The Strength and Difficulties Questionnaire (SDQ) is recommended for screening for social/emotional problems in children aged 3 to 13 (Ruiter & Jong, 2013). This is a simple tool to use and is freely available (www.sdqinfo.org).
- The Four Dimensional Symptoms List (4DSL) or Depression Recognition Scale (DRS) are recommended for screening for social/emotional problems in adolescents and adults (Ruiter & Jong, 2010).

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Chapter 10: How should Proper Aftercare be Organised and Implemented?

10.1 Introduction

Stuttering is a tricky problem. Improvement in speech with the various techniques and/or attitude changes already described is perhaps easily achievable in the clinical setting, however applying all of this and maintaining improvement in daily practice is often difficult. Recurrence occurs frequently, and significant benefits may only be achieved if the PWS is able to internalise the transfer of therapy in non-clinical situations (i.e. during participation in society); the PWS has to become his/her own therapist. Aftercare is of crucial importance in this process. It was apparent from the Focus Group meeting that the form of aftercare has not been adequately defined. Both the Dutch Stutter Association Demosthenes and the Dutch Association for Stutter Therapy (NVST) believe that aftercare requires a programmatic approach. The concepts aftercare and self-help need to be clearly distinguished from each other in this process.

10.2 Method

No systematic literature reviews have been undertaken in order to answer this principal question. The report from the PWS focus group meeting (CBO, 2013) was used for the description of the patient perspective. For the description of the professional perspective use has been made of the knowledge, experience and opinions of the working group members acting as speech & language therapists/fluency specialists.

Definitions of aftercare and self-help

The concepts aftercare and self-help are not always clearly distinguished from each other in the literature about stuttering. In this guideline the working group's understanding of aftercare is taken from the definition in Van Dale, the Dutch dictionary, as "the pathway following the end of the pursued therapy". The operationalisation of this depends on the individual circumstances of the PWS or the child who stutters (CWS). The initiative for contact may come from either the PWS or from the speech & language therapist or fluency specialist. The Dutch Council for Healthcare Quality defines aftercare as "*The entirety of measures aimed at returning a patient to an as normal a situation as possible, with as much independence as possible. Aftercare may comprise physical, psychological and societal measures*".

In the context of this guideline the working group understands aftercare as: the pathway following the end of therapy with the associated care and aftercare appointments. The abovementioned definition provides a direction in which a speech & language therapist or fluency specialist should organise an aftercare programme.

For clarification of the distinction between aftercare and self-help, the working group will follow the Dutch Council's definition of "self-help": "*A form of help provision based on the principle that people are able to help themselves and each other by being aware of, and exchanging similar experiences, as well as through participating in activities*" (Dutch Council for Healthcare Quality, 2013). In terms of self-help, the terms "self-help/mutual aid group" and "support group" are not always strictly distinguished in the international literature. The existing self-help groups, and for instance, the "stutter cafes" supported by the Dutch Stutter Association Demosthenes are relevant for self-help in the Netherlands.

Programmatic approach: definition and elements

In its "Advice regarding Aftercare in Oncology", the Dutch Health Council has argued for "*bundling written information into an aftercare plan for each patient at the end of treatment, and for this to be made available for the patient, GP and other parties concerned. An aftercare plan such as this contains more than just the aftercare programme. At the very least it contains information about the physical and psychosocial consequences of the disease and treatment, the desirability and design of the aftercare review, and a moment of reflection and consideration of any remaining focus points*".

Good quality aftercare

The aims of good quality aftercare were defined with a scheme (IOM, 2001) in the Oncology Care Guideline (www.oncoline.nl/dimensies, dated 2 November 2013). The working group has adopted that scheme to describe good quality aftercare in stuttering. The principles of this scheme may be used by the speech & language therapist or fluency specialist in drawing up an aftercare plan for stuttering.

Principles of good aftercare in stuttering

Quality dimension	Patient-focused quality	Professional quality	Organisational quality
Good aftercare for stuttering	promotes quality of life, daily functioning and the patient's self-sufficiency	is based on the most up-to-date insights from science (evidence) and the profession (expert opinion)	is integrated within speech & language therapy for stuttering as a structural component of the care continuum
	is differentiated care, linking in to the needs (physical, psychological, social) and preferences of the patient (tailored care)	is offered with the best level of expertise	is accessible for each patient
	is based on autonomy and self-determination of the patient and on his/her active participation	is offered in/with open communication	is coordinated care with a permanent contact for the patient
	is respectful	is targeted and effective	is safe

10.3 Evidence

Recurrences in former stuttering patterns are regularly reported. This is apparent from, amongst other things, a survey conducted with (adult) members of the National Stuttering Association (NSA), with the aim of exploring the experiences with support groups or support groups in combination with speech & language therapy (Yaruss et al., 2002).

Recurrence occurs for all age groups and underlines the significant relevance of focusing on embedding aftercare within the therapy process. Proper aftercare satisfies the needs of the PWS - even if this is not experienced explicitly in the first instance (Bezemer, Bouwen & Winkelman, 2010; Manning, 2010).

The PWS's environment needs to be involved at an early stage in the treatment; this should preferably occur at the start of therapy, but should certainly be indicated in the transfer, the maintenance phase and aftercare.

Aftercare organised in the form of an "open end" therapy may be pleasant during the final stages of therapy 'the door is ajar' as it were (Focus Group Report, meeting 17 April 2013).

Peer group contact (including, amongst others, the Dutch Stutter Association, Demosthenes) is a very valuable experience, particularly for adolescents and adults; the realisation that there are others with comparable problems may have a significant enlightening effect (Boyle, 2013).

Contact such as this with peers may form a component of therapy (particularly in group therapy), but peer contact may also be very useful at the end of therapy (for instance, in the form of self-groups and stutter cafes). Whilst participation in groups such as these is encouraged and, in principle is an unlimited resource, it may be useful for the participant to draw up a plan of action and goals for this. Secondary factors (for instance, travel distance) may be relevant here.

Tailored aftercare, organised and shaped directly after therapy, prepared in a timely fashion, and available for two years after therapy, satisfies the needs of the PWS and is of importance in managing stuttering.

10.4 From Evidence to Recommendations

10.4.1 Desired and Undesirable Effects of Aftercare versus Self-Help

The speech & language therapist/fluency specialist views aftercare as an emphatic part of therapy. However, it is difficult to realise in clinical practise. This appears to be associated with a reduced degree of ill-health, cost-savings in healthcare and the absence of a clear aftercare framework for everyone (Klink, 2013).

Participation in a support group may be care-related (aftercare guided by a care professional), and it may also be a form of self-help. A potential undesired effect for the PWS may be that this distinction is not always clear. Insurance companies contribute to this lack of clarity by reimbursing some forms of group sessions without the intervention of a professional, relevantly trained healthcare provider. When a PWS is considering looking for help in the form of group sessions it is advisable that he/she has sufficient information in advance; utilisation of the advice published by the International Stuttering Association (ISA) in respect to the questions that need to be posed to the therapist is recommended.

http://www.isastutter.org/wp-content/uploads/2013/06/Declaration-for-Stuttering-Treatment.pdf

10.4.2 Professional and Patient Perspectives

The speech & language therapist / fluency specialist takes a methodological, stepwise approach to therapy; a part of this is aftercare. A programmatic approach to aftercare in the form of an aftercare plan falls under the umbrella of quality care for stuttering. The speech & language therapist/ fluency specialist should establish tailored and focused support as the basis for aftercare during therapy. This can be multidisciplinary: sometimes (temporary) support is necessary to link in the needs (physical, psychological, social) and preferences of the PWS. The focus on patient self-management is highly relevant as part of the (after)care of patients and is of increasing interest in healthcare (Engels & Kistenmaker 2009).

In addition to this aftercare may be organised in the context of self-help or through tele-health (e.g. http://www.stotters.nl/) – the PWS has his/her own virtual therapist, who may sometimes be an extension of his/her own therapist.

According to the Oncology Care Guideline (www.oncoline.nl) there has been little specific research in the Netherlands or Europe into the way in which aftercare can be optimally organised; however, individualisation is certainly an important aspect to this. Patients require aftercare that fits their situation and want to be able to contribute to decisions (Cardella et al., 2007). The medical choices should be made by the specialist in consultation with the patient (Milliat-Guittard et al., 2007).

10.4.3 Costs and Aftercare

The exact figures about the costs of aftercare following treatment for stuttering are not known. Peer group contact often occurs voluntarily at a minimal cost.

It is plausible that an inadequately planned trajectory of aftercare increases the risk of recurrence. This means, in many cases, that the PWS will enter into another therapy trajectory again over the course of time, which of course will incur costs. Although the costs of aftercare are not clear, it is logical that these are less than new treatment.

Recommendation

25. Any therapeutic approach to stuttering must include methods which promote long-term effects, and deal with possible setbacks. As a part of the therapy process, the way in which follow up support is organized, is defined by the SLT and the PWS in collaboration with his environment. At the end of the clinical treatment the SLT proposes a programmed approach for an individualized follow up program lasting for 2 years. In order to prevent relapse and to promote long-term effects, the SLT suggests the PWS and his environment to contact other PWS, e.g. activities organized by patients' associations

Rationale for the recommendations

- Recurrence occurs frequently, and significant benefits may only be achieved if the PWS has internalised the transfer of therapy into non-clinical situations (i.e. during participation in society) and has an aftercare trajectory at hand.
- The concepts aftercare and self-help are not always clearly distinguished from each other in the literaturearound stuttering.
- The aims of good quality aftercare need to be defined.
- In the literature on stuttering and in clinical practice the usual, desirable duration of aftercare is two years.
- The working group was unable to establish any accurate estimates on the costs of aftercare in drafting these recommendations, mainly through the lack of information about current costs.

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Appendix 1: Study Assessment

Assessor's name: Hans de Beer. Date: 11-02-2013

Title: Stuttering Treatment Research 1970-2005 (I +II). Authors: Bothe et al. Source: Am J of Speech-language Pathology 15 (2006).

Research question: To complete a systematic review, incorporating a trial quality assessment, of published research about behavioural, cognitive (and related), pharmacological treatments for stuttering. Goals included the identification of treatment recommendations and research needs based on the available high-quality evidence.

Validity assessment:
METHODS

1. Was the research question properly formulated?
[X] Yes
[ ] No
[ ] Too little information in the article to answer this

2. Was the search conducted properly?
[X] Yes (comment: no search in Cinahl, some key words are missing such as *stammering* or *dysfluency*)
[ ] No
[ ] Too little information in the article to answer this

3. Was the selection procedure for the articles carried out properly?
[ ] Yes
[X] No (describe how the selection procedure was not carried out properly? By 1 or 2 people?)
[ ] Too little information in the article to answer this

4. Was the quality assessment carried out properly?
[X] Yes (although not entirely; for instance it was not very transparent)
[ ] No
[ ] Too little information in the article to answer this

5. Was there an adequate description of how the data extraction was undertaken?
[X] Yes
[ ] No
[ ] Too little information in the article to answer this

6. Have the most important features of the original studies been described?
[X] Yes (but only moderately, as interventions and patient groups have not been described in detail)
[ ] No
[ ] Too little information in the article to answer this

7. Has the clinical and statistical heterogeneity in the studies been handled properly?
[ ] Yes
[ ] No
[X] Too little information in the article to answer this

8. Has statistical pooling been conducted in the correct manner?

[X] Yes / not applicable (no pooling was carried, which is correct due to the heterogeneity)

[ ] No

[ ] Too little information in the article to answer this

Conclusion: Reasonably good systematic review.
Categorisation of the methodological quality of systematic reviews:
Score of 7-8 (7-8 x yes answers): good
Score of 6: reasonably good
Score of ≤5: moderate

Assessor's name: Hans de Beer. Date: 19-03-2013
Title: Effectiveness of Behavioral Stuttering Treatment: A Systematic Review and Meta-Analysis
Authors: Herder et al. Source: CONTEMPORARY ISSUES IN COMMUNICATION SCIENCE AND DISORDERS •
Volume 33 • 61–73 • Spring 2006

Research question: The purpose of the present study was to conduct a systematic review and meta-analysis of the effectiveness of behavioural stuttering treatments for people who stutter.

Validity assessment:

Brief description of the patient categories: those who had been diagnosed as PWS using labels such as stuttering or stammering.

Brief description of the intervention(s) assessed:

All included studies used a behaviourally-based intervention as the method to improve speech behaviours.

Brief description of the control treatment(s):

All included studies identified both experimental and control (or comparison) groups to which participants were assigned to before the intervention. A control group is defined as a non-treatment condition; a comparison group is defined as an alternative treatment condition.

METHODS
1. Was the research question properly formulated?

[X] Yes

[ ] No

[ ] Too little information in the article to answer this

2. Was the search conducted properly?

[X] Yes

[ ] No

[ ] Too little information in the article to answer this

3. Was the selection procedure for the articles carried out properly?

[X] Yes

[ ] No

[ ] Too little information in the article to answer this

4. Was the quality assessment carried out properly?

[ ] Yes

[X] No

[ ] Too little information in the article to answer this

5. Was there an adequate description of how the data extraction was undertaken?
[ ] Yes
[X] No
[ ] Too little information in the article to answer this

6. Have the most important features of the original studies been described?
[ ] Yes
[X] No
[ ] Too little information in the article to answer this

7. Has the clinical and statistical heterogeneity in the studies been handled properly?
[X] Yes
[ ] No
[ ] Too little information in the article to answer this

8. Has statistical pooling been conducted in the correct manner?
[X] Yes / not applicable
[ ] No
[ ] Too little information in the article to answer this

Conclusion:
Moderate quality systematic review
Categorisation of the methodological quality of systematic reviews:
Score of 7-8 (7-8 x yes answers): good
Score of 6: reasonably good
Score of ≤5: moderate

Appendix 2: Evidence Table for Effectiveness of Behavioural Interventions

Study (1st author, year of publication)	Randomisation adequate?	Allocation concealed?	Groups similar at baseline?	Patient blinded?	Care provider blinded?	Outcome assessor blinded?	Cointervention avoided/similar?	Compliance acceptable?	Drop-out rate described/acceptable?	Timing outcome assessment similar?	Intention to treat analysis?	No selective reporting of outcome?	Risk of bias per study per outcome measure
Perkins 1974 (N=44;	-	-	-	NA	NA	-	?	?	-	+	+	+	Speech tempo: *high* Stutter frequency: *high* SEC-variables: *high*
James 1989 (N=24; 4 drop-outs)	-	-	-	NA	NA	-	?	?	±	+	+	+	Speech tempo: *high* Stutter frequency: *high*
Ladouceur 1986 (N=16)	-	-	?	NA	NA	-	?	?	?	+	+	+	Speech tempo: *high* Stutter frequency: *high*
Miltenberger 1996 (N=2)	-	-	-	NA	NA	-	?	?	+	+	+	+	Speech tempo: *high* Stutter frequency: *high*
Saint-Laurant 1987 (N=40)	+	-	?	NA	NA	+	+	?	+	-	+	+	Speech tempo: *moderate* Stutter frequency: *moderate*
Ost 1976 (N=15)	±	-	?	NA	NA	+	+	?	+	+	+	+	Speech tempo: *moderate* Stutter frequency: *moderate*
Carey et al (2010)	+	+	+	NA	NA	+	+	+	+	+	+	? (speech tempo not evaluated)	Stutter percentage: *low*
Cream et al (2010)	+	+	+	NA	+	+	+	-	+	+	+	+	All outcomes (see text): *low to moderate (due to limited compliance)*

| Study (1st author, year of publication)	Randomisation adequate?	Allocation concealed?	Groups similar at baseline?	Patient blinded?	Care provider blinded?	Outcome assessor blinded?	Cointerventions avoided/similar?	Compliance acceptable?	Drop-out rate described/acceptable?	Timing outcome assessment similar?	Intention to treat analysis?	No selective reporting of outcome?	Risk of bias per study per outcome measure
Menzies et al (2008)	+	?	-	NA	NA	+	+	?	- (>20%)	+	-	+	Stutter frequency: moderate Social anxiety: moderate
Huinck et al (2004)	-	-	-	NA	NA	?	+	+	-	+	-	+	For all outcome measures: moderate/high

Appendix 3: Evidence Table for Behavioural and Cognitive Interventions

Author(s): Hans de Beer. Date: 2013-03-06

Question: Should behavioural and cognitive approaches be used for stuttering?

Bibliography: Bothe et al. Stuttering Treatment Research 1970-2005: I. Systematic Review Incorporating Trial Quality Assessment of Behavioral, Cognitive, and Related Approaches, American Journal of Speech-Language Therapy, 15, 321-341.

Quality assessment							No of patients		Effect		Quality	Importance
No of studies	Design	Risk of bias	Inconsistency	Indirectness	Imprecision	Other considerations	Behavioural and cognitive approaches	Control	Relative (95% CI)	Absolute		
Stuttered syllables (follow-up >=6 months; Better indicated by higher values)												
6	randomised trials[1]	serious[2]	serious[3]	no serious indirectness	no serious imprecision[4]	None	141[5]	-	-	not pooled	⊕⊕⊝⊝ LOW	CRITICAL
Number of syllables per minute (follow-up >=6 months; Better indicated by lower values)												
6	randomised trials[1]	serious[2]	serious[3]	no serious indirectness	no serious imprecision[4]	None	141[5]	-	-	not pooled	⊕⊕⊝⊝ LOW	CRITICAL

[1] Not all studies were randomised trials, but there were no observational studies either.

[2] Most studies were not randomised. The outcomes were not ascertained blind in the majority of studies. There is uncertainty about the comparability of groups.

[3] There was considerable variance in outcomes for the individual studies.

[4] This is difficult to determine as the data are lacking to produce a combined estimate.

[5] This relates to the total number of evaluated trial participants.

Appendix 4: Overview of Search Actions for Stuttering

File Name	Number
med 20120730 speech and language therapy guidelines na 1995	13*
med 20120731 aanvulling speech and language therapy guidelines na 1995	13*
psy 20120731 speech and language therapy guidelines na 1995	33*
cl systrev 20120808 speech and language systrev	13
med 20120730 speech and language therapy systrev na 1995	111
psy 20120808 speech and language developmental systrev na 1995	29
emb 20120808 speech and language therapy guidelines vanaf 1995	64
emb 20120808 speech and language therapy systrev vanaf 1995	11
cin 20120809 speech guidelines	10*
cin 20120809 speech systematic reviews	37
Update van 12 feb 2013	
med 20130212 stotteren systrev na 1995	2
med 20130212 stotteren rct na 1995	99
psy 20130212 stotteren systrev trials na 1995	107
emb 20130212 P stotteren trials systrev	94
cin 20130212 P stotteren systrev	6
cin 20130212 P stotteren trials	10

* These were not easily usable in this guideline.

Search action of 20120730
Database: Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) <1946 to Present>

Search Strategy:
----
1 "stotteren richtlijnen".ti. (0)
2 exp "rehabilitation of speech and language disorders"/ (7997)
3 guideline/ or practice guideline/ (22758)
4 (speech adj therap*).ti. (549)
5 Speech Therapy/ (4799)
6 Language Therapy/ (1105)
7 4 or 5 or 6 (5602)
8 guidelin*.ti. (42983)
9 3 or 8 (56465)
10 7 and 9 (22)
11 10 (22)
12 limit 11 to yr="1995 -Current" (14)
13 2 and 9 (22)
14 13 (22)
15 limit 14 to yr="1995 -Current" (13)
16 exp Stuttering/ (2891)
17 stutter*.tw. (3251)
18 stammer*.tw. (265)

19 fluency disorders.mp. (41)
20 dysfluen*.tw. (213)
21 non-fluen*.tw. (285)
22 (language adj3 thera*).tw. (1201)
23 (speech adj therap*).tw. (1926)
24 2 or 5 or 6 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 (13404)
25 9 and 24 (39)
26 25 not 10 (17)
27 26 (17)
28 limit 27 to yr="1995 -Current" (14)
29 from 15 keep 1-13 (13)
30 from 28 keep 1-14 (14)
31 (dutch or english or french or german).la. (18765055)
32 24 and 31 (12119)
33 32 (12119)
34 limit 33 to yr="1995 -Current" (6136)
35 "med091027 CBO filter sysrev & meta Medline START".ti. (0)
36 meta analysis.pt. (35060)
37 (meta-anal$ or metaanal$).af. (62641)
38 (quantitativ$ adj10 (review$ or overview$)).tw. (3692)
39 (systematic$ adj10 (review$ or overview$)).tw. (43338)
40 (methodologic$ adj10 (review$ or overview$)).tw. (5233)
41 medline.tw. and review.pt. (33113)
42 (pooled adj3 analy*).tw. (6029)
43 or/36-42 (117710)
44 "med091027 CBO filter sysrev & meta Medline EINDE".ti. (0)
45 34 and 43 (142)
46 (language adj thera*).tw. (975)
47 (speech adj therap*).tw. (1926)
48 4 or 5 or 6 or 16 or 17 or 18 or 19 or 20 or 21 or 46 or 47 (10784)
49 31 and 48 (9846)
50 49 (9846)
51 limit 50 to yr="1995 -Current" (4941)
52 43 and 51 (115)

■■

Search of 20130212
Database: Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) <1946 to Present>

Search Strategy:
----
1 "ontwikkelings stotteren opbouw zoekactie".ti. (0)
2 (speech adj therap*).ti. (561)
3 Speech Therapy/ (4871)
4 Language Therapy/ (1134)
5 exp Stuttering/ (2918)
6 stutter*.tw. (3282)
7 stammer*.tw. (267)
8 fluency disorders.mp. (41)
9 dysfluen*.tw. (214)
10 non-fluen*.tw. (290)
11 (language adj3 thera*).tw. (1257)
12 (speech adj therap*).tw. (1985)
13 or/2-12 (11147)

14 "P voor stotteren".ti. (0)
15 guidelin*.ti. (44111)
16 guideline/ or practice guideline/ (23179)
17 (dutch or english or french or german).la. (18999648)
18 13 and (15 or 16) and 17 (38)
19 limit 18 to yr="1995 -Current" (28)
35 "med091027 CBO filter sysrev & meta Medline START".ti. (0)
21 meta analysis.pt. (36967)
22 (meta-anal$ or metaanal$).af. (66218)
23 (quantitativ$ adj10 (review$ or overview$)).tw. (3867)
24 (systematic$ adj10 (review$ or overview$)).tw. (47091)
25 (methodologic$ adj10 (review$ or overview$)).tw. (5405)
26 medline.tw. and review.pt. (34357)
27 (pooled adj3 analy*).tw. (6385)
28 or/21-27 (124498)
29 "med091027 CBO filter sysrev & meta Medline EINDE".ti. (0)
30 13 and 17 and 28 (151)
31 limit 30 to yr="1995 -Current" (143)
32 developmental.ti. (40859)
33 developmental.tw. (166702)
34 Developmental Disabilities/ (13319)
35 speech disorders/ or stuttering/ (12206)
36 35 and (33 or 34) (660)
37 ((stutter* or fluency* or stammer* or clutter*) adj3 developmental).tw. (159)
38 (dysfluen* adj3 developmental).tw. (6)
<t score="N/A"> (435)
40 (d?sfluen* adj3 developmental).tw. (13)
41 (early adj stutter*).tw. (47)
42 or/36-41 (1091)
43 13 and (33 or 34) (604)
44 42 or 43 (1408)
45 (child??? or childhood or infant* or p?ediatr* or perinat* or neonat* or newborn* or infan* or boy? or girl? or kid? or schoolage* or juvenil* or adolescen* or toddler?).tw. (1558274)
46 exp Child/ (1456397)
47 exp infant/ (886722)
48 "Adolescent"/ (1501278)
49 45 or 46 or 47 or 48 (3084391)
50 "filter child cbo medline".tw. (0)
51 44 and 49 (1287)
52 51 and 17 (1192)
53 limit 52 to yr="1980 -Current" (1050)
54 "P voor stotteren bij kinderen".ti. (0)
55 language therapy/ or speech therapy/ (5562)
56 (th or rh).fs. (1471365)
57 (intervent* or therap*).ti. (593790)
58 treat*.ti. (960849)
59 or/55-58 (2561520)
60 53 and 59 (442)
61 "med101005 Cochrane Highly Sensitive Search Strategy for Randomized Trials in Medline START".ti. (0)
62 randomized controlled trial.pt. (339011)
63 controlled clinical trial.pt. (85097)
64 (randomized or randomised).ab. (306310)
65 placebo.ab. (140242)
66 drug therapy.fs. (1573096)
67 randomly.ab. (187872)

68 trial.ab. (264547)
69 groups.ab. (1216413)
70 or/62-69 (3045624)
71 70 not (exp animals/ not humans/) (2604016)
72 "med101005 Cochrane Highly Sensitive Search Strategy for Randomized Trials in Medline EINDE".ti. (0)
73 60 and 71 (99)
74 from 19 keep 1-28 (28)
75 from 31 keep 1-140 (140)
76 from 60 keep 1-3 (3)
77 from 73 keep 1-96 (96)
78 "bothe$".fc_auts. and "2006".fc_pubyr. and "321".fc_pg. (1)
79 exp *Stuttering/ (2646)
80 stutter*.ti. (2694)
81 79 or 78 or 7 or 8 or 9 or 10 (3204)
82 2 or 3 or 4 or exp Stuttering/th or 11 or 12 (7942)
83 81 and 82 and 17 (914)
84 83 and (15 or 16) (3)
85 (81 or 82) and 17 (9301)
86 85 (9301)
87 limit 86 to yr="1995 -Current" (4839)
88 87 and (15 or 16) (26)
89 87 and 28 (134)
90 5 or 6 or 7 or 8 or 9 or 10 (4286)
91 89 and 90 (21) med 20130212 stotteren systrev na 1995
92 87 and 71 and 90 (387)
93 "med091027 CBO filter rct Medline START".ti. (0)
94 randomized-controlled-trial.pt. (339011)
95 controlled-clinical-trial.pt. (85097)
96 randomized controlled trial/ (339011)
97 randomi?ed controlled trial?.tw. (64762)
98 random-allocation.af. (77042)
99 double-blind-method.af. (117486)
100 single-blind-method.af. (16981)
101 (random adj8 (selection? or sample?)).tw. (30915)
102 random$.tw. (625670)
103 or/94-102 (846073)
104 "med091027 CBO filter rct Medline EINDE".ti. (0)
105 controlled clinical trial/ or randomized controlled trial/ (419357)
106 from 91 keep 1-21 (21)
107 Clinical Trials as Topic/ (162088)
108 103 or 105 or 107 (958898)
109 87 and 90 and 108 (101) med 20130212 stotteren rct na 1995

Appendix 5: Definitions and Concepts

Confidence Interval

There is always a degree of uncertainty in research. This is because a restricted group of patients are studied in order to predict the effects of treatment for a larger population. The confidence interval is a way of expressing how certain we are about the findings in a study. The confidence interval is usually described as "95% CI", which means that the range of values has 95 out of 100 chance of containing the "true" value. For instance, a study may note that "based on our findings we're 95% certain that the "true" blood pressure of the population is not higher than 150 and not lower than 110". In that the 95% CI would be 110-150. A wide confidence interval indicates a lack of certainty about the actual effect of the test or treatment - often because a small group of patients has been studied. A narrow confidence interval provides a more accurate estimate (for instance when a large number of patients has been considered).

Blinding

This is a way of preventing researchers, doctors, paramedics and patients in a clinical trial from knowing which study group a patient belongs to, so they cannot influence the results. The best way to do this is to assign patients at random to study groups. The aim of "blinding" is to prevent or limit bias. A single-blinded study is a study in which patients do not know to which study group they belong (for instance whether they are taking the experimental medicine or a placebo). A double-blinded study is a study in which neither the patients nor the researchers/doctors know to which study group the patients belong. A triple-blinded study is a study in which neither the patients, the clinicians nor the people undertaking the statistical analysis know which treatments the patients have undergone.

Case Report

Non-controlled observational research involving only 1 or a handful of patients.

Cinahl

Cumulative Index of Nursing and Allied Health Literature. This database, published by Cinahl Information Systems, California (U.S.), includes references to publications from 1982 to the present time in the field of nursing and professions allied to health. The database may only be consulted through subscription. The CBO has a subscription. Information about Cinahl may be found out: http://www.cinahl.com .

Cochrane Collaboration

An international organisation with the aim of offering support to informed decision-making processes in healthcare. This is achieved through publishing systematic reviews and meta-analyses about the effects of healthcare interventions.

Cochrane Library

A collection of databases from the Cochrane Collaboration, including the most important databases: a) Cochrane Database of Systematic Reviews, the most recent summaries of which are also in PubMed and b) Database of Abstracts of Reviews of Effectiveness (DARE), these are systematic reviews, that are not Cochrane reviews, which have been evaluated on quality.

Effect Size

A measure representing the size of the outcome in a group in comparison with a control group. For instance, if the absolute risk reduction is 5% and this is an outcome in which researchers are interested, then the effect size is 5%.

The effect size is usually tested using a statistical test in order to assess what the chance is that the effect results from the treatment and not due to chance alone (that is to determine whether it is statistically significant).

Concealment of Allocation

In an RCT people in the study population are randomly allocated (for instance using sealed envelopes) into intervention group(s) and control group(s). "Concealment of allocation" refers to the process of keeping secret or blinding this allocation of patients from the various study groups. This means that the individual allocating the groups (for instance by handing out envelopes) is not aware of the contents of the envelope and that coding cannot be uncovered either.

Heterogeneity

This term is used in meta-analyses and systematic reviews to indicate that the results of a test or treatment (in other words effect sizes) differ significantly across various studies. These differences may occur as a result of differences in the population investigated, the outcomes measured or due to differences in definitions for the variables in question.

Inconsistency (synonym: substantial heterogeneity)

Inconsistency reflects unexplained heterogeneity in the results, in other words highly variable results for the treatment effect.

Indirectness

The biomedical literature is not able to answer the principal question due to differences in patients, interventions, control groups or outcome measures. Indirect comparisons are also an example of indirectness, for instance comparing the outcomes from intervention A with those from intervention B on the basis of intervention A versus placebo and intervention B versus placebo.

Intention-to-Treat Analysis

An assessment of the outcomes in participants in a clinical trial based on the group they were randomly allocated to in the first instance. This is irrespective of whether they dropped out, completed the treatment in full or switched to a different treatment group. Intention-to-treat analyses are used to determine clinical effectiveness, because they reflect current practice: that is, not everyone adheres to treatment and treatment is adapted in accordance with how people react to it.

Core Stutters or Core Stuttering Behaviour

These are the tense repetitions, extensions and blockades of syllables and sounds that are characteristic of stuttering. The disruptions to the fluency of speech are involuntary and the person who stutters does not have them under control.

Clinical Significance

A beneficial effect of a treatment that is related to an important outcome, such as survival, and is large enough to be important for patients and care professionals. Effects that are statistically significant are not always clinically significant, where the effect is small or the outcome measures are not important. For instance, if a treatment increases blood flow, but there is no evidence that this leads to an important clinical outcome, such as a smaller risk of a heart attack, then there is no evidence of clinical significance.

Quality of Evidence (also described as *certainty in respect to the effect size*; in line with the GRADE definition)

Quality of evidence reflects the degree of certainty about an effect.

High quality of evidence: further scientific research will not change the confidence in the *effect estimate;*

Moderate quality of evidence: further scientific research will *probably* have an important impact on our confidence in the effect estimate, and *may* change the *estimate;*

Poor quality of evidence: further scientific research is very likely to have an important impact on our confidence in the effect estimate and will *probably* change the *estimate*;

Very poor quality of evidence: *all estimates of the* effect are *uncertain*.

Quality of Life

Is the functioning of individuals in the physical, psychological and social domains and the subjective evaluation associated with this? Quality of life therefore consists of both relatively objective, as well as subjective aspects. Objective conditions concern the fact of whether someone has limitations as a result of his/her health. Subjective aspects say something about the person's judgement of (aspects of) his/her health (Sprangers MAG (AMC)). What is quality of life and how is it measured? In: Volksgezondheid Toekomst Verkenning, Nationaal Kompas Volksgezondheid (Exploring the Future of Public Health, the National Public Health Monitor). Bilthoven: RIVM, <http://www.nationaalkompas.nl> Nationaal Kompas 2009.)

Medline

A database with references of articles from about 5000 journals, published in more than 70 countries. The majority of the published articles are from English-language journals. The search language is in English. The database extends back to approximately 1950 and contains more than 14 million references. Medline may be consulted for free via PubMed: http://www.ncbi.nlm.nih.gov/sites/entrez . The CBO has a subscription to a more advanced search interface for Medline.

Meta-analysis

A statistical analysis of the results of independent studies with the aim of providing a summary estimate of the effect (of a treatment). A meta-analysis is often part of a systematic review where the results of a number of comparable clinical studies have been bundled together. This enables a judgement to be made about the effect of an intervention or treatment with a greater degree of confidence.

Non-comparative Research

Research which does not involve a comparison of groups; this includes descriptive studies and case reports.

Naturalness of Speech

Is the degree to which the speech of a PWS sounds like that of a typical speaker who does not stutter. (Guitar 2014)

Odds Ratio

Odds are a way of showing how likely it is that something will happen (the likelihood). An odds ratio compares the likelihood of something in a group compared to the likelihood of the same thing in a different group.

An odds ratio of 1 between two groups would indicate that the likelihood of the event (for instance whether the therapy is effective) is the same in both groups. An odds ratio greater than 1 means that the event is more likely in the first group. An odds ratio smaller than 1 means that the event is less likely in the first group. Sometimes the likelihood of an event may be compared between more than two groups - in this event one of the groups is chosen as the "reference category", and the odds ratio is calculated for each group in comparison with the reference category. For instance, in order to calculate the mortality risk for lung cancer in non-smokers, casual smokers and habitual smokers, non-smokers may be used as the reference category. Odds ratios could be calculated for the casual smokers compared to the non-smokers and for the habitual smokers compared to non-smokers.

Inaccuracy

Inaccuracy means uncertainty about the effect size, as reflected by a wide confidence interval, and is caused by a limited number of people investigated in a study.

Developmental Stuttering

The most common form of stuttering which develops during childhood (as opposed to stuttering that develops as a result of a neurological condition, a trauma or through emotional stress) (Guitar, 2014).

PICO-method

This acronym describes four elements (Patient, Intervention, Control, Outcome) of a specific clinical question which can be used for a search in the scientific literature.

PsycINFO

A database with references to articles in psychology, created by the American Psychological Association and containing descriptions and abstracts of articles, book chapters, books and theses in the field of psychology and the psychological aspects of all related disciplines. Data originate from 1500 journals from 1872. The previous name of the database was PsycLit. The database may only be consulted with a subscription.

PubMed

PubMed is a database with descriptions (references) of journal articles from biomedical, nursing and dental journals. It contains more than just Medline. It contains PubMed, indexed for Medline, PubMed as supplied by the publisher and PubMed in Process; these are references included in Medline, but that have not yet been provided with MeSH terms (Medline keywords). Pubmed is freely accessible via: http://www.ncbi.nlm.nih.gov/sites/entrez. See also medline.

P-value

The p-value or threshold value (of a given sample outcome) is the probability of achieving or exceeding (left, right or two-sided) the observed test result given that the null hypothesis is true. For instance, if a study is comparing two treatments, and one appears to be more effective than the other, the p-value is the likelihood of these results arising by chance alone. The convention is that a p-value less than 0.05 (or a probability of less than 5% of these results arising by chance) is considered as a value below which it is likely that there is an actual difference between both treatments. The confidence interval describes how large this difference in effect could be.

Randomised Controlled/Clinical Trial (RCT)

This is research where the effect of an intervention is compared with that of a control intervention and with non-selective allocation (randomisation) of patients into the index group and reference group. RCTs are considered to be the best research methods to test a hypothesis concerning medical interventions. In a placebo-controlled RCT the control group will be administered a placebo.

Risk of Bias

Synonym: systematic error.

When bias arises the results will differ from the true effect due to a systematic error. Bias may occur as a result of flaws in the design of the study, in collecting the data, in analysing and interpreting the results and in publishing. Bias, however, cannot always be avoided. Selection bias occurs when the distortion of the results is caused by an essential difference in individuals included in and excluded from the study. For instance, during the inclusion process systematically selecting those individuals for whom the intervention studied will have the greatest effect.

Allocation bias (a form of selection bias) whereby the participants in a study are not allocated non-selectively to the study groups, for instance due to incorrect randomisation procedures. Information bias refers to an error arising in the measurement of the parameters investigated (also known as information bias, measurement bias, or assessment bias). The fault may lie with the researcher, for instance when no clear definitions of the research parameters have

been established (interviewer bias, observer bias, or interpretation bias). The patient may also contribute to bias by not being able to remember an important fact (recall bias).

Response bias is when some people agree with certain statements, irrespective of the contents or when respondents only provide socially desirable answers.

Confounding bias is bias that arises as a result of the fact that insufficient data was taken of the disruptive effect of confounders on the relationship between the central determinant and the disease. A confounder is a disruptive variable responsible for a biased representation of the relationship between the central determinant and the disease. Another form of bias is publication bias. If negative outcomes lead to certain study results not being published then there is a publication bias. This form of bias is important in the assessment of systematic reviews and meta-analyses.

Secondary Stuttering Behaviour

Is the result of a (partly unconscious) learning process by the person who stutters as a reaction to the core stuttering behaviours. Examples include flight and fight behaviours, such as blinking, loss of eye contact, physical movements, tongue movements, the use of run-up sentences ("erm..erm..what's it called..."), etc. Other reactions include feelings of anxiety, frustration and shame and avoidance behaviours (in words and/or sounds that the person who stutters uses and in situations).

Stuttering Severity

Is a measure based on the objective measurement of stuttering behaviour, such as stuttering frequency, duration of stutter moments, the type of dysfluencies and secondary behaviours (Shapiro, 1999).

Stuttering-Like Dysfluencies (SLDs)

A type of dysflueny in young children which help to distinguish "normal dysfluency" from "stuttering" (Yairi & Ambrose, 2005). These dysfluencies occur much more frequently in children who stutter:
- repeating parts of words
- repeating single letter group words
- dysrhythmic phonations, such as tense pauses

% Stuttered Syllables (%SS)

The percentage of stuttered letter groups is a measure of the stuttering frequency.

Systematic Review

A systematic literature review of the state-of-affairs of medical/scientific research concerning a specific question. A systematic review is transparent and reproducible and is based on an explicit question, an extensive search strategy, a transparent procedure for selection of the

studies, a systematic evaluation of the quality of the studies and a comprehensive presentation of the results. A meta-analysis may be part of a systematic review. Refer also to meta-analysis.

Systematic Literature Review
Refer to systematic review

Principal Question
This is a specific (clinical) question for which an answer will be provided in the guideline in the form of a recommendation. The principal questions form the basis for the development of the guideline and the starting point for the literature review.

Outcome Measure
That what is used to measures the result of an event or intervention. A distinction is made between hard endpoints, such as death or evident morbidity, and intermediary or surrogate endpoints, which are usually only indirectly associated with the hard endpoints. Examples include the serum lipid levels and blood pressure, where the occurrence of cardiovascular disease is the hard outcome measure. Effects of interventions on intermediary endpoints cannot be easily extrapolated to effects on hard endpoints. The value of studies that only provide intermediary endpoints is therefore limited.

Appendix 6: Constraints Analysis - Stuttering Guideline

The CBO in collaboration with the Dutch Association for Logopedics and Phoniatrics (NVLF), the Dutch Association for Stutter Therapy, and the patient association "Demosthenes" has developed an evidence-based guideline for stuttering in children and adults. The guideline is intended to describe what the diagnosis, treatment and aftercare of clients who stutter should look like. Professional organisations indirectly involved in the diagnosis and treatment of clients who stutter were approached to participate in a focus group. The associations participating in the focus group include:

- NHG Netherlands Society of General Practitioners
- AJN Netherlands Association of Paediatricians
- NIP Netherlands Institute of Psychologists
- NVK Netherlands Paediatric Society
- KH Netherlands Society of Otolaryngology and Head and Neck Surgery
- NVAB Netherlands Society of Occupational Medicine
- LBBO National Occupational Group of Special Needs Education Professionals

Method

A survey was created in order to detail the constraints around the diagnosis, treatment and aftercare of stuttering in children and adults. This survey was sent to the associations represented in the working group, which included speech & language therapists, fluency specialists and patients, and to those associations represented in the focus group. A total of 40 surveys were completed and returned. Eight surveys were completed during uni-disciplinary meeting of which six surveys originated from the NVST fluency specialists, one from the Paediatric Physicians Association and one from the Dutch Association for Work and Business Studies. One survey was completed by the board of the patient association, Demosthenes. In order to obtain a different set of views, ten surveys were completed by members of the NVLF, ten by members of the NVST, eight by members of the Demosthenes patient association, one by a GP representative from the Dutch Family Practitioners Association, one by a clinical psychologist, and one by a paediatrician from the Dutch Association for Paediatric Medicine.

An overview of the answers submitted may be found in the file "results stutter survey". A review of the results will be described in this report.

A maximum of 8 research questions will be formulated following the constraints analysis.

Review of the Surveys

It is particularly evident from the surveys that there is a need for greater clarity around the detection, referral, treatment and aftercare in stuttering. It is important to create greater clarity about when the point of diagnosis needs to take place, the contents of the diagnosis, the content and timing of treatments and when and to whom the patient who stutters should be referred.

Below the points that were highlighted in the survey will be discussed per subject.

Internal Constraints

The survey highlighted the constraints in terms of the diagnosis, treatment and aftercare that occur in daily practice in respect to the care of children and adults who stutter. These constraints may be an important source for formulating the research questions. Given that referral is often experienced as a constraint, referrals have been included as a separate component in this constraints analysis.

Constraints Analysis

The point at which an organisation takes action and makes a referral when suspecting stuttering is frequently experienced as a constraint. The Paediatric Physicians Association indicated they were not sufficiently aware of the signs that indicate the need for referral. Patients and speech & language therapists/fluency specialist also indicated that the moment at which a referral is made to a speech & language therapist/fluency specialist, psychologist or other organisations after treatment has had little success and is regularly too late.

Diagnosis

Constraints are experienced regarding which diagnostics to apply and the associated time required for diagnosis; there is also a debate around who should be conducting the diagnosis. Patients, speech & language therapists/fluency specialists indicated that stuttering is a multifactorial problem requiring more than just diagnosis and treatment of speech. They have their doubts in this process whether the sole practitioner is the right professional to tackle the full extent of the stuttering problems, to set the right priorities in terms of the aspects on which the intervention should be focused and the point at which the intervention should start.

Treatment

Stuttering is described in the literature as a multifactorial communication problem, often associated with comorbidity in terms of language, articulation, sensory motor skills, cognitions, and emotions and behaviour; these problems come to the fore in the social/communicative context. The mutual exchange of the various factors is difficult to pin down.

There are different options available to treat stuttering. It is not well known which intervention is the most effective for individual patients who stutter. Speech & language therapists/fluency specialists indicated that protocols and guidelines based on proven effectiveness are not available to determine the steps to be taken and which goals to set. This is experienced as an even greater problem if there is comorbidity. A unilateral emphasis on speech and, especially, fluent speech was described by fluency specialists, paediatricians and members of the patient association as too limited and potentially harmful. There are question marks about who is best able to implement treatment or certain aspects of treatment. Variability and multi-causality of stuttering demands a great deal more research and agreement with the parties concerned compared to other speech & language problems. Speech & language therapists/fluency specialists indicated that the financing is inadequately regulated to provide the client with the optimum level of care required.

Aftercare

Recurrence following therapy for stuttering has been described in the literature on numerous occasions. It is clear from the survey that speech & language therapists, fluency specialists and patients view aftercare as an integral part of treatment. It is not known or written in detail how the content, timing and duration of aftercare should be organised.

Constraints in the Organisation and the Process of Care

The guideline for stuttering in children and adults offers the opportunity to optimise the collaboration between the various specialities involved in the care of people who stutter through joint agreements about the organisation of care, collaboration and communication.

Organisation of Care

Children and adults with developmental stuttering are often referred too late for extensive diagnosis. On the one hand there is frequently a delayed referral by GPs, paediatricians and other referring organisations. On the other hand parents and teachers acknowledge that further research is required. This is possibly due to insufficient knowledge about the incidence, course of stuttering and insufficient awareness of the opportunity for referral, but it is also due to stuttering sometimes being minimalised: "It will go away". The guideline for stuttering in children and adults may generate improvements in terms of the chain of detection and referral by parents, teachers, GPs, paediatricians to speech & language therapists, fluency specialists, psychologists or a multidisciplinary diagnostic team.

Agreement between the various disciplines during the diagnostic and intervention phases appears to be limited. This agreement could be improved by a clear division of work and promoting mutual collaboration.

Constraints were experienced in terms of detecting and referring, division of work in care, tailoring the care and financing the care.

Collaboration and Communication

The guideline for stuttering in children and adults may generate good initiatives for adequate detection of potential stuttering by parents and healthcare providers and a more rapid referral by doctors. It is of importance here that communication about stuttering and the treatment of the development of stuttering is improved across all parties concerned. Proper agreement about the detection, referral, diagnosis, treatment, guidance and aftercare between the client and/or parents of the client, speech & language therapist, fluency specialist, GPs, paediatricians, ENT physicians, psychologists, paediatric physiotherapists, teachers and playgroup leaders is essential. Clear collaboration based on protocols should lead to enhanced communication.

Prioritisation

A number of themes were highlighted in the prioritisation of the constraints which can be tackled in the stuttering guideline:

- Emphasising the importance of timely detection and correct diagnosis of stuttering.
- The importance of extensive diagnosis by experts who not only assess the speech motor aspect, but also the stuttering problem in its entirety.
- Determining the optimal treatment, including aftercare, for the individual client on the basis of the best available scientific evidence with a focus on the total person in his/her social/emotional context.
- Design a clear pathway of care from the detection to aftercare with a clear overview of consultation and referral options with other disciplines when associated problems are detected that are hindering treatment.
- Communication about stuttering and the treatment options provided to the laity and healthcare professionals.
- Financing in accordance with the care that needs to be provided.

Appendix 7: Abbreviations

AAF
Altered Auditory Feedback

BAB
Behaviour Assessment Battery

CWS
Child Who Stutters

DCM
Demands and Capacities Model

ICF
International Classification of Functioning, Disabilities and Health

LP
Lidcombe Program

%SS
Percentage stuttered syllables

OASES
Overall Assessment of Speaker's Experience of Stuttering

PWS
Person who stutters

TfS-R/NR
Test for stuttering severity readers/non-readers (TvS: Test voor Stotterernst)

RCT
Randomised Controlled Trial

SSI
Stuttering Severity Instrument

SSL
Stuttering Screening List (SLS: Screenings Lijst Stotteren)

SLT
Speech and Language Therapist