Evidence-based Clinical Guidelines in Stuttering Therapy

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Topics

• Aims and definition.
• The Dutch Evidence-based Clinical Guideline Developmental Stuttering:
  – developmental proces,
  – grading the evidence,
  – from evidence to recommendations.
• Guidelines in Europe.
• Which therapy?
• When to start therapy?
• When to end therapy?
• Who has to provide therapy?
Undesirable Variation
Evidence-based Clinical Guidelines

• Reduce undesirable variation,
• Improve transparency,
• Make the rapidly growing flow of information manageable,
• Integrate clinical expertise with the best available scientific evidence.
The Ultimate Goal

Improve quality of care!
Definition

“An Evidence-based guideline is a document with recommendations meant to improve quality of care. It is founded on systematic reviews of scientific research and on the assessment of the advantages and disadvantages of different care options and includes expertise and experience of healthcare professionals and service users.”

(Richtlijn voor Richtlijnen, 2011)
Evidence-based guidelines can be used to formulate standards of care.

Standards of care describe the process of care necessary to provide good healthcare.
The Dutch Evidence-based Guideline Developmental Stuttering in Children and Adults
Uncertainty about..

Content of Care

- Diagnostics
- Intervention

Organisation of Care

- Timing
- Refering
- Collaboration
- Aftercare
Available Stuttering Guidelines

• Guidelines for Practice in Stuttering Treatment
  American Speech and Hearing Association
  (1995)

• Clinical Guidelines
  Royal College of Speech & Language
  Therapists (2005)
Available Stuttering Guidelines

- The most influential guidelines are developed nationally and interpreted locally (Barkham et al. 2010)

- Update every 5 years
The Working Group

- Bast, dr. E.J.E.G., Demosthenes
- Bezemer M., NVST
- Beer, dr. ir. J.J.A. de, CBO
- Bunschoten, E.M., NVST
- Kalter, E., MSc., NVLF
- Kuijpers, dr. A.C., CBO
- Oonk, L.C., MSc., chair
- Ormondt, J. van, Demosthenes
- Pertijs, M.A.J., MSc., project manager
- Rosenbrand, drs. C.J.G.M., CBO
- Wijngaarden, van, L.J., VSN
Advisory and Focus Group

The advisory group
- Family doctors
- Specialists
- Teachers
- Psychologists

Focus group
- Persons Who Stutter
- Parents from Children Who Stutter
Evidence-based Guideline Development

- Define search criteria
  - databases
  - time period
  - PICO

- Literature search
- Define selection criteria
- Selection of literature

- Formulation of key questions

- Problem analysis
  - Medical
  - Organizational
  - Patient issues

- Implementation feedback

- Dissemination & publication
- Finalize guideline + endorsement by scientific medical associations

- Re-writing draft guideline
- External review
  - National open meeting
  - Scientific medical associations/societies
  - Website

- Finalize draft version of guideline
- Development of indicators
- Re-writing draft

- Critical appraisal of selected literature
- Evidence table
  - diagnostics
  - therapy

- Writing first draft of guideline
  - Summary statement of the evidence
  - Further considerations
  - Recommendations for clinical practice

- Discussion of draft guideline
  - subgroup
  - planair
Key Questions

1. What are the indications to treat children and adults with developmental stuttering?
2. What is the value of diagnostic test (I1, I2, ...) in children and adults with developmental stuttering?
3. What are the positive and negative effects (O1, O2, ...) of a particular treatment (I1, I2, ...) compared with no treatment or with another treatment (C1, C2) in children and adults with developmental stuttering (P)?
Key Questions

3. When and for what reason should a PWS be referred to a specific care provider (e.g. SLT, Stuttering Specialist, Psychologist, etc..)?

4. How should adequate aftercare be organized and implemented?
Grading the Evidence
Clinical Guidelines
Royal College of Speech & Language Therapists (2005)

<table>
<thead>
<tr>
<th>Level Type of evidence (based on AHCPR 1992)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ia Evidence obtained from meta-analysis of randomised controlled trials</td>
</tr>
<tr>
<td>Ib Evidence obtained from at least one randomised controlled trial</td>
</tr>
<tr>
<td>IIa Evidence obtained from at least one well-designed controlled trial without randomisation</td>
</tr>
<tr>
<td>IIb Evidence obtained from at least one other type of well-designed quasi-experimental study</td>
</tr>
<tr>
<td>III Evidence obtained from well-designed non-experimental descriptive studies, such as comparative studies, correlation</td>
</tr>
<tr>
<td>IV Evidence obtained from expert committee reports or opinions and/or clinical experience of respected authorities</td>
</tr>
</tbody>
</table>

Bias
**Grade Recommendations (based on AHCPR 1994)**

**A** Requires at least one randomised-controlled trial as part of the body of literature, of overall good (Evidence levels Ia, Ib) quality and consistency, addressing the specific recommendation.

**B** Requires availability of well-conducted clinical studies but no randomised clinical trials on the (Evidence levels IIa, IIb, III) topic of recommendation.

**C** Requires evidence from expert committee reports on opinions and/or clinical experience of (Evidence level IV) respected authorities. Indicates absence of directly applicable studies of good quality.
1) Demands and Capacities Model (B)

These therapy programmes are based on the principle that a child's capacity for fluency can be enhanced naturally by reducing the internal and external demands that may be being placed on a vulnerable system. Learning two languages does not necessarily increase a child's risk of stammering.

**Rationale**

Stammering is a highly variable, context-sensitive disorder. Most children who stammer have the capacity to be more fluent at certain times according to a range of internal and external factors, within differing communicative environments.

**Evidence**

- Guitar B, Schaefer HK, Donahue-Kilburg G, & Bond L (1992) *Evidence Level III*
- Weiss AL & Zebrowski RM (1992) *Evidence Level III*
- Gottwald SR (1999) *Evidence Level IV*
- Yalni E (1997) *Evidence Level IV*

2) Child (fluency) focused therapy (A)

These therapies aim at direct modification of the child's stammering, usually involving a behavioural methodology and, in some cases, involving parents/carers. In the Lidcombe Program the therapist's role is to teach the parents how to carry out the therapy at home.

**Rationale**

Early stammering is viewed as a maladaptive response that can be replaced by more adaptive behaviours through the use of appropriate and timely feedback. Such therapies need to be carefully structured.

**Evidence**

- Onslow M, Andrews C, & Lincoln M (1994) *Evidence Level IIa*
AHCP-R-System

Limitations

• confuse quality of evidence with strength of recommendations
• criteria not comprehensive or transparent
• lack well-articulated conceptual framework
Grading the Evidence

GRADE

Grades of Recommendation Assessment, Development and Evaluation
GRADE is outcome-centric

Old system

GRADE

Outcome #1

Outcome #2

Outcome #3

Quality: High

Quality: Moderate

Quality: Low

Kuijpers & De Beer (2013)
GRADE is outcome-centric

Old system

Patient Important Outcomes

Outcome #1
Outcome #2
Outcome #3

GRADE

Quality: High
Quality: Moderate
Quality: Low
Rates of Relative Importance of Outcomes

- Critical for making a decision
- Important, but not critical for making a decision
- Not important
<table>
<thead>
<tr>
<th>Outcome Measures</th>
<th>Relevance (patients perspective)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stuttering severity</td>
<td>critical</td>
</tr>
<tr>
<td>Avoidance behaviour</td>
<td>critical</td>
</tr>
<tr>
<td>Speech naturalness</td>
<td>important</td>
</tr>
<tr>
<td>Quality of life</td>
<td>critical</td>
</tr>
</tbody>
</table>
## Quality criteria

<table>
<thead>
<tr>
<th>Design</th>
<th>Initial quality of a body of evidence</th>
<th>Lower if</th>
<th>Higher if</th>
<th>Quality of body of evidence</th>
</tr>
</thead>
<tbody>
<tr>
<td>RCT</td>
<td>High $\rightarrow$</td>
<td>Study limitations</td>
<td>Large effect</td>
<td>High</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Inconsistency</td>
<td>Dose response</td>
<td>Moderate</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Indirectness</td>
<td>All plausible confounding &amp; bias</td>
<td>Low</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Imprecision</td>
<td>- would reduce a demonstrated effect</td>
<td>Very low</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Publication bias</td>
<td>- would suggest a spurious effect when results show no effect</td>
<td></td>
</tr>
<tr>
<td>Observational study</td>
<td>Low $\rightarrow$</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
## Quality of Evidence (4 categories)

The quality of evidence reflects the extent of our confidence that the estimates of the effect are correct.

<table>
<thead>
<tr>
<th>Quality</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>High</td>
<td>We are <strong>very</strong> confident that the true effect lies close to that of the estimate of the effect.</td>
</tr>
<tr>
<td>Moderate</td>
<td>We are <strong>moderately</strong> confident in the effect estimate: The true effect is likely to be close to the estimate of the effect, but there is a possibility that it is substantially different.</td>
</tr>
<tr>
<td>Low</td>
<td>Our confidence in the effect estimate is <strong>limited</strong>: The true effect may be substantially different from the estimate of the effect.</td>
</tr>
<tr>
<td>Very Low</td>
<td>We have <strong>very little</strong> confidence in the effect estimate: The true effect is likely to be substantially different from the estimate of effect.</td>
</tr>
</tbody>
</table>

Kuijpers & De Beer (2013)
Key Question

What are the desirable and undesirable effects of treatment according to the Demands and Capacities Model compared to treatment with the Lidcombe Program on stuttering severity, avoidance behaviour, speech naturalness and quality of life in CWS 2 to 6 years of age?
Quality of Evidence Lidcombe Program (PICO)

<table>
<thead>
<tr>
<th>Study</th>
<th>Treatment n</th>
<th>Control n</th>
<th>Hedges’ g</th>
<th>Lower limit CI</th>
<th>Upper limit CI</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Harris et al. (2002)</td>
<td>8</td>
<td>11</td>
<td>0.67</td>
<td>-0.23</td>
<td>1.56</td>
<td>.144</td>
</tr>
<tr>
<td>Jones et al. (2005)</td>
<td>27</td>
<td>20</td>
<td>0.94</td>
<td>0.34</td>
<td>1.54</td>
<td>.002</td>
</tr>
<tr>
<td>Latterman et al. (2008)</td>
<td>23</td>
<td>22</td>
<td>1.51</td>
<td>0.53</td>
<td>1.77</td>
<td>.001</td>
</tr>
<tr>
<td>Overall Effect</td>
<td></td>
<td></td>
<td>0.97</td>
<td>0.58</td>
<td>1.30</td>
<td>.001</td>
</tr>
</tbody>
</table>

Effect size, 95% Confidence Interval and p-value of percentage stuttered syllables comparing children 2 to 6 years of age for treatment vs control group RCT design with post-test measurement immediately following intervention. (Nye et al., 2012).
## Quality of Evidence Lidcombe Program (PICO)

<table>
<thead>
<tr>
<th>Quality of evidence:</th>
<th>Treatment of stuttering children under 6 years with the Lidcombe Program in the short and medium term (3 to 9 months) is more effective in reducing the percentage of stuttered syllables than when stuttering is not treated. Nye et al (2012)</th>
</tr>
</thead>
</table>

The observed effect can be considered large ($g = 0.97$, 95% CI: 0.58 to 1.30, $p <.0001$). Effects larger than 0.7 are considered large (Higgins & Green, 2008). We are **moderately** confident in the effect estimate. This implies that there is some uncertainty about the magnitude of the effect. The effect could also be 0.58.
### Quality of Evidence DCM (PICO)

<table>
<thead>
<tr>
<th>Quality of evidence:</th>
<th>The number of studies of treatment based on DCM is limited and our confidence in the effect estimate of these studies is limited. The true effect might be substantially different from the estimate of the effect.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Low</td>
<td>No clinically relevant difference in the reduction of the %SS was demonstrated after treatment comparing DCM and the Lidcombe Program group. However, there is considerable uncertainty about this conclusion, given the width of the 95% confidence interval (( g = -0.275^*, 95%CI: -1.066 - 0.517 )) in the study by Franken et al. (2005).</td>
</tr>
</tbody>
</table>

*Threshold clinical relevant difference in reduction \%SS is (-)0.5 according to the GRADE working group.*
Balance of Desired and Undesired Effects

- The most commonly reported outcome measure is % SS, other outcomes are hardly or not reported.
Balance of Desired and Undesired Effects

• The effect of treating CWS up to 6 years with the Lidcombe Program in the short and medium term (3 to 9 months) is large compared with no treatment.

• Over the long-term effects of treatment with the LP exists greater uncertainty.
Balance of Desired and Undesired Effects

- No evidence that proved undesirable effects of treatment with the Lidcombe Program.

<table>
<thead>
<tr>
<th>Quality of evidence:</th>
<th>There are no indications of possible adverse effects of the Lidcombe Program such as anxiety, aggression, avoidance and depression and the impact on the quality of the mother and child relationship.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Very Low</td>
<td>Woods et al. (2002)</td>
</tr>
</tbody>
</table>
Balance of Desired and Undesired Effects

- Effect of treatment with DCM shows a substantially similar magnitude of effect compared to the effect size of treatment with the LP.
- Undesirable effects of DCM are hardly investigated or reported.
Values and Preferences

Parents of CWS

• Inform parents about the possible benefits of treatment.

• Preferences of parents play an important role in the decision to choose treatment with DCM or the LP.

SLT’s and Fluency Specialists

• Become competent in treating CWS with the LP

• It is an advantage to become competent in treating CWS with DCM as well.
Inform parents of children who stutter under 6 years of age about the benefits of treating stuttering compared to no treatment*. In consultation with the parents of the young child who stutters decide which method is preferred, the Lidcombe Program or treatment based on the Demands and Capacities Model.

*The recommendation about timing of therapy (starting therapy or monitoring) will be described at the recommendation no.1 (indications to treat CWS).
GRADE

• conceptual framework
• comprehensive, transparent criteria
• focus on all important outcomes related to a specific question and overall quality
## Undesirable Variation in Europe

<table>
<thead>
<tr>
<th>Country</th>
<th>Clinical Guideline</th>
<th>Type</th>
</tr>
</thead>
<tbody>
<tr>
<td>Austria</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Belgium</td>
<td>No*</td>
<td></td>
</tr>
<tr>
<td>Croatia</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Cyprus</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Finland</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>France</td>
<td>?</td>
<td></td>
</tr>
<tr>
<td>Germany</td>
<td>Yes</td>
<td>Consensus based, Ev-B under construction</td>
</tr>
<tr>
<td>Greece</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Ireland</td>
<td>Under construction</td>
<td>Evidence-based</td>
</tr>
<tr>
<td>Italy</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Macedonia</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Malta</td>
<td>Yes</td>
<td>Evidence-based</td>
</tr>
<tr>
<td>Norway</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Poland</td>
<td>Standard</td>
<td>Consensus based</td>
</tr>
<tr>
<td>Portugal</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Slovenia</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>Spain</td>
<td>?</td>
<td></td>
</tr>
<tr>
<td>Sweden</td>
<td>Under construction</td>
<td>Evidence-based</td>
</tr>
<tr>
<td>The Netherlands</td>
<td>Under construction</td>
<td>Evidence-based</td>
</tr>
<tr>
<td>United Kingdom</td>
<td>Yes</td>
<td>Evidence-based</td>
</tr>
</tbody>
</table>
Evidence-based Guideline Development
The Evidence-based Guideline Developmental Stuttering is sponsored by:

- De Nederlandse Vereniging voor Logopedie en Foniatrie,
- Het Damsté-Terpstra Fonds,
- De Nederlandse Vereniging voor Stottertherapie,
- De Nederlandse Stottervereniging Demosthenes.