The German interdisciplinary evidence- and consensus-based guideline on the pathogenesis, assessment, and treatment of speech fluency disorders: What is internationally generalizable?

http://www.awmf.org/leitlinien/detail/ll/049-013.html

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Why a guideline?
• In order to properly identify, diagnose and treat speech disfluency disorders, evidence-based German guidelines on stuttering and cluttering have been elaborated and published in September 2016.
• It was necessary to review the current knowledge on fluency disorders, because it has changed during the past years, in particular with respect to the causes of stuttering, and its neuro-structural and neuro-physiological underpinnings.
• To change the view was in particular important because there is a strong psychoanalytical tradition in Germany, Austria, and Switzerland with an ideographic approach to stuttering.
• Furthermore, a recent study (Euler et al. 2014) showed which treatments of stuttering in Germany are effective and which are not.
• Patients have the right to receive the treatment with the best evidence, hence therapists need to know and apply these treatments.

Where can you find the guideline?

http://www.awmf.org/leitlinien/detail/ll/049-013.html

https://www.aerzteblatt.de/int/archive/article?id=189154

Classification of Speech Fluency Disorders

Because the originary neurogenic stuttering, which arises during childhood with no recognizable causes, is associated with structural and functional changes in the brain we propose to change the attribution of the word 'neurogenic' with respect to stuttering.

Definition
• Because stuttering symptoms do not belong to the normal language development and because stutter typical disfluencies may be differentiated from normal speech disfluencies we propose not to use the term 'developmental stuttering'.
• The genuine neurogenic non-syndromal stuttering is a central nervous speech and speech planning disorder which arises during childhood due to a genetic disposition. It comprises core symptoms with stutter-typical disfluencies and a secondary symptoms with vegetative, motor, and emotional reactions on the speech disfluency disorder.

Epidemiology
• Genuine neurogenic non-syndromal stuttering starts between 2 to 6 years of age in most cases
• Prevalence:
  - children and adolescents (2 to 18 yrs.) 1.4 %
  - adults: <1 %
• Sex distribution:
  - in the initial phase: >3:2 male : female
  - later, due to sex-different recovery 5:2 male : female
• Incidence:
  - up to 4th/5th yrs. 5.0 % - 11.2 % (depending on study) due to high recovery rate  much higher than prevalence
Recovery

- Sponanteous recovery: in 70-80 % of cases
- Regarded permanent if speech fluency kept >12 mos.
- Occurs mostly until puberty, rare in adulthood
- Recovery rate is highest in the first 2 yrs., in particular during the first 6-12 mos.
- Thereafter, chances for a recovery decrease drastically
- Risk factors for persistence:
  - male
  - stuttering persists >2y
  - persistent stuttering in family
  - age of stuttering onset >4yrs.
  - symptoms increased in severity
- However, an individual prognosis regarding recovery cannot be made.

Stuttering symptoms

- Origininy neurogenic non-syndromal stuttering has to be diagnosed, if at least 3 % of syllables in a representative speech sample show stutter typical disfluencies (repetitions of sounds, syllables, and monosyllabic words, prolongations, blocks)
- Independent from the stuttering frequency (i.e. also if <3 % of stuttered syllables are there) stuttering has to be assumed and diagnostics has to be performed in case of long-lasting stuttering events, emotional load by the stuttering, avoidance behaviors, and other secondary symptoms such as strain in the stutter symptom or stutter-associated co-movements.

German manner of counting stuttered and non-stuttered syllables: from polysyllabic whole-word or utterance repetitions, revisions, and interjections. 'Ich bin gestern, also gestern, ahem, gestern bin i - ich hier gewesen.' includes in the 17 spoken syllables 2 stuttered and 15 non-stuttered syllables, independent whether the interjections ('also gestern', 'ahem') are interpreted as avoidance behavior or not (this kind of counting is described in the manual of the SG-4 (Stuttering Severity Instrument, 4th edition, Riley 2000).

Genetics of stuttering

- The originary neurogenic non-syndromal stuttering is highly heritable (70-80 %).
- Molecular genetics has identified more than a dozen disposition loci for stuttering on chromosomes 1, 2, 3, 5, 7, 9, 12, 13, 15, 16, 18, and 21.
- One locus on chromosome 8 has been identified, the allel of which is shared by all persons who do not stutter (protective factor?)
- The language input for a child is not involved in the primary causes of stuttering.
- By counselling parents, speculations about the potential causal role of the socialization of a child with respect to her stuttering have to be avoided.
- Parents shall be reassured that they are not responsible for the occurrence of their child's stuttering.

Cerebral structural and functional correlates of stuttering

- Origininy neurological stuttering is associated with predominantly genetically caused cerebral morphological and functional abnormalities compared with non-stuttering persons, in particular in circuits, which are involved in speech, language, and auditory processes.
- The production of speech fluency needs a continuous dynamic interaction between auditory, somatosensory, and speech motor neuronal networks.
- According to the dual-stream-model of the cerebral language architecture (Hickok & Poeppel 2004, 2007), in stuttering - which is regarded predominantly as a disturbance of the speech motor control and less a linguistic problem – in particular dorsal language circuits are disturbed which are involved in the auditory-motor integration. This reflects a disturbed processing of internal feedforward mechanisms (projections of the motor plans, which are sent to the sensory system for perception according to a planned movement) and auditory feedback mechanisms (GODIVA-Model), paralleled by a disturbed fitting of the heard own speech in the speech-motor planning and execution. After successful therapy of stuttering adults, a reduction of excessive activity in dorsal language regions has been observed.

Comorbidities of stuttering

- Social anxiety
- Language factors like phonological, semantic-lexical or grammar abilities do not seem to play a major role for children at kindergarten ages (2-6 years) for the occurrence and persistence of stuttering
- If a superiority or inferiority of stuttering compared with non stuttering children with respect to their language development and communicative abilities have been found, the deviations were small in both directions.
- Slightly higher coincidence with learning disabilities and dyslexia?
- Attention deficit disorder?
- Less stuttering in children with deafness or profound hearing loss?

All named comorbidities are rather questionable!
Diagnostics of stuttering according to the ICF model

Diagnostics of stuttering shall assess
the core symptoms and accompanying behaviors (body function), the psycho-social load (person-related factors), communication & social behavior (activity & participation) & reactions from the social environment (environmental factors), objective (quantity & quality of core & motor behavior), perceptual measures as evaluated by others (description of symptoms, accompanying behavior, speech naturalness) and by the patient herself (subjective perspectives of the patient, consequences for the everyday life, and health-related quality of life).

Functioning and Disability
- Body functions & structures
- activity & participation
- environmental factors
- person-related factors
- context factors

Diagnostics of stuttering

The following objective measurements of the audible and visible stuttering symptoms shall used:
• Stuttering Severity Instrument (SSI-4, Riley 2009), applicable for all age groups from 2;10 years on
• Test of Childhood Stuttering (TOCS, Gillam et al. 2009) , applicable for 4- to 12-year old
• The OASES (Overall Assessment of the Speakers’ Experience with Stuttering, Yaruss & Quesal 2006, 2008, Yaruss, Quesal et al. 2010, Yaruss, Coleman et al. 2010) or a comparable but shorter German questionnaire (Fragebogen zur psychosozialen Belastung durch das Stottern für Kinder und Jugendliche, Cook 2013) shall be applied in order to assess the health-related quality of life in the age groups, for which age-related normative values are available.

If there is a suspicion of an additional psychic disorder patients need to undergo a psychological/psychiatric diagnostics according to the state of the art.

Screening for stuttering

At-risk screenings: for children who are suspect for stuttering or have higher probability, for example because of stuttering relatives
• Recommendable: Screening List for Stuttering (SLS, Riley & Riley 1989)

Universal screenings: for example general language screenings during regular pediatric examinations at ages 3, 4, and 5 yrs. (in Germany), or school entry screenings
• At current (in Germany): during regular pediatric examinations at ages 3, 4, and 5 yrs. parents are asked by examiners whether their child stutters, and speech is evaluated
• Recommendable: Aachener Stotterscreening (BASS, Neumann et al. 2014), German tool for physicians, suited for general language screenings during regular pediatric examinations, language screenings in kindergartens, or school entry screenings

Diagnostics of stuttering

Stuttering and bi-/multilingualism

• Bilingual children should not be kept away from the early access to a second language because parents are afraid of a potential stuttering.
• Stuttering of a bi-/multilingual person may be differ in extent between the different languages she speaks.
• Usually, a treatment of stuttering in more than one language is not necessary.

Diagnostics of stuttering

Stutter ratings

• Ratings of speech naturalness should be performed by non-professional examiners, if possible by averaging the scores of several examiners.
• Ratings of stuttering severity should be performed by third professionals who are not involved in the treatment.
• Global ratings of the stuttering severity shall not be used as the only measure of the treatment effect, if they are done by the therapist herself.
As always, a systematic review is generally better than an individual study. Inclusion criteria for treatment reports

- Informs about treatments aimed at reduction of stuttering
- Sample size ≥1
- Effect sizes reported or calculable
- At least 2 within-subject repeated measures
- Follow-up ≥3 months after end of treatment; pre-post alone insufficient

→ 43 studies included in the systematic review

**Classification of levels of evidence acc. to Oxford Centre for Evidence-Based Medicine 2011 - Levels of Evidence**

* Level may be graded down on the basis of study quality, imprecision, indirectness (study PICO does not match questions PICO), because of inconsistency between studies, or because the absolute effect size is very small; Level may be graded up if there is a large or very large effect size.

** As always, a systematic review is generally better than an individual study.

**Tables of Evidence**

<table>
<thead>
<tr>
<th>Evidence</th>
<th>Level of evidence</th>
<th>Judgement of study quality</th>
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<tbody>
<tr>
<td>Lidcombe</td>
<td>++ high</td>
<td>++**</td>
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<tr>
<td><strong>(1) Lidcombe treatment</strong> (verbal reinforcement of fluent utterances by parents, gentle correction for disfluent utterances) Strong evidence of efficacy (10 high quality studies) Strong recommendation: &quot;is to be applied&quot;</td>
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<tr>
<td><strong>(2) Indirect methods</strong> (parents trained to adjust their speech to the capabilities of the child) Strong evidence of efficacy for the Dutch RESTART-DCM method 1 (high quality study) Recommendation: &quot;ought to be applied&quot;</td>
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<td>Treatment of preschool children (≤ 6 yrs)</td>
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**Evidence for Lidcombe program**

Authors | Level of evidence | Judgement of study quality |
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<tr>
<td>Nye et al. (2013)</td>
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<td>Bothe et al. (2006a)</td>
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<td>Jones et al. (2005)</td>
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<td>De Sonnevile-Koedoot et al. (2015)</td>
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<td>Lattermann et al. (2008)</td>
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<td>Jones et al. (2000)</td>
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**Strong evidence of efficacy** (10 high quality studies) Strong recommendation: "is to be applied"
### Evidence for indirect methods

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<th>Studienqualität</th>
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<tr>
<td>1</td>
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<td>31</td>
<td>Franken et al. (2005)</td>
<td>Pilotstudie</td>
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### Treatment of preschool children (≤ 6 yrs)

Insufficient evidence of effectiveness for most popular treatment:
**Extensive unspecific treatment** (1 session/week; e.g. speaking, reading, breathing practices) by SLP (logopedist) in private practice or school setting.

Recommendation:
Require quantitative baseline assessment. *If no improvement after 3 months, reconsider.*

### Treatment of school children (6 to 12 yrs)

Absent, poor, insufficient or negative evidence for any treatment method

Treatment gap for this age group

Indication, that children of this age may profit from speech restructuring or a combination of speech restructuring and stuttering modification

### Treatment of adolescents and adults (≥12 yrs)

1. **Speech restructuring** (e.g. Fluency Shaping, Campervan, Slowed Speech): Patient is taught a new speech pattern which avoids disfluencies.

   *Strong evidence of efficacy (11 high quality studies)*

   *Strong recommendation: “is to be considered”*

2. **Stuttering modification**: Patient is taught techniques to overcome local blocks.

   *Weak evidence of efficacy (3 high quality studies): Open recommendation: *may be considered*

3. **Combination of Speech restructuring and Stuttering modification**

   *Weak evidence of efficacy (4 high quality studies): Open recommendation: “may be considered”*

### Evidence for Speech restructuring

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<td>23</td>
<td>Bothe et al. (2006a)</td>
<td>für Kinder 6-12 Jahre keine Empfehlung möglich, aber gute Empfehlung für Erwachsene</td>
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### Evidence for Stuttering modification

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<td>7</td>
<td>Euler et al. [2014]</td>
<td>Einschätzung der Wirkung durch Patienten</td>
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<td>18</td>
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<td>33</td>
<td>Blomgren et al. (2005)</td>
<td>neg. Empfehlung für SMP</td>
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<td>36</td>
<td>Laiho &amp; Klipp (2007)</td>
<td>6</td>
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### Evidence for combination from speech restructuring and Stuttering modification

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Treatment of adolescents and adults (≥12 yrs)

(4) Pharmacological agents

Insufficient evidence of efficacy (2 high quality studies)

Strong recommendation: “shall not be administered”

Other, less evidenced treatments:

- Gradual Increase in Length and Complexity of Utterance (GILCU): may be considered
- Extended Length of Utterance (ELU): may be dispensed with
- Rhythmic speaking: negative evidence; ought not be used as sole treatment component
- Breathing regulation: negative evidence; ought not be used as sole treatment component
- Hypnosis: weak negative evidence; ought not be applied
- Unspecified stuttering treatments: weak negative evidence; ought not be applied
- Acceptance and Commitment Therapy (ACT): ought to be dispensed with
- Speech Motor Training: ought to be dispensed with

Cluttering

- For cluttering, the authors of the guideline found less evidence with respect to a pathophysiologically based definition, (assessment,) and in particular treatment.
- Neuroimaging experiments have shown cerebral alterations of the speech processing of persons who clutter compared with both normally fluent persons and studdering persons (Neumann et al. 2015; Ward et al. 2015)
- Hence, cluttering has to be regarded an own pathogenetic entity (and not a norm variant).
- Research is needed to further elucidate this disease.

Drafting committee

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Thanks to all the contributors

and

Thank you for your attention!