Recovery from Stuttering

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- By **Peter Howell**
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- Description
- Contents
- Subjects
- **Description**
- This book is a comprehensive guide to the evidence, theories, and practical issues associated with recovery from stuttering in early childhood and into adolescence. It examines evidence that stuttering is associated with a range of biological factors—such as genetics—and psychological factors—such as anxiety, and it critically assesses theoretical accounts that attempt to integrate these findings. Written so that it can be used flexibly to meet the demands of courses about stuttering, the book may be used as a text at the undergraduate or graduate level in psychology or speech-language science.

Recovery is "taboo"
- **Because people claim to be able to "cure" stuttering**
- **Mainly**
    - **drugs**
    - **by alteration to voice feedback**
    - **behavioural treatments like CBT**
- **All of these are examples of "treatment assisted"**

Recovery in young children allowed.
- **Called "natural recovery"**
- **Not clear how to distinguish the two forms of recovery**
- **Critiques:**
- **Natural – no recourse to professional advice, else it is treatment assisted**
- **Treatment assisted – not ruled out natural recovery rate, so natural recovery could be a part of supposed natural recovery**

Definition of recovery and plan of talk:

1. State (stuttering)

2. Change/no change of state (recover/persist)

3. Process of change

Topic one, the state of stuttering

Stuttering is difficult to define

An early list of symptoms that occurred frequently in children who stutter (Johnson et al., 1959):

1) Incomplete phrases (sometimes called abandonments)

2) Revisions (change of one word for another)

3) Interjections (word and non-word fillers)

4) Whole-word repetitions ("I, I, I")

5) Phrase repetitions ("in the, in the morning")

6) Part-word repetitions ("k-k-Katy")

7) Prolongations ("sssister")

8) Broken words ("di-nosaur")

Observations:

1) Acknowledge that all shown by fluent speakers

2) Many drop abandonments, revisions, interjections (1-3)

3) Opinion split about 50/50 concerning whether to included whole-word repetitions

Slide 1

- **Authors who do include:**
- Bernstein (1981)
- Bloodstein and Gantwerk (1967)
- Bloodstein and Grossman (1981)
- Conture (1990)
- Reilly et al. (2009)
- Wingate (1964)
- Yairi and Ambrose (2005).

- **Authors who do not include**
- Bernstein Ratner and Sih (1987)
- Gaines, Runyan and Meyers (1991)
- Dejoy and Gregory (1985)
- Jayaram (1981)
- Kloth et al. (1999)
- Riley (1994)
- Wall, Starkweather and Harris (1981)
- Wingate (1988; 2002)

Slide 2

Issue 1 Status of whole-word repetitions

Debate has been seen as whether to include or exclude whole-word repetitions.

Later we will consider an alternative position (that whole-word repetitions have a different role to more typical stuttered disfluencies)

Wingate's three arguments to exclude whole-word repetitions:

1) They occur in the speech of all speakers
2) They happen naturally in the speech of fluent speakers so they are not barriers to producing speech fluently
3) These symptoms occur in the speech of speakers who stutter and there is no compelling evidence that they differ in any way from the same symptoms produced by fluent speakers.

Slide 3

Evidence whether to include whole-word repetitions

1) **Anderson (2007)** Anderson (2007) found that the word, and neighborhood, frequency of words that had a part-word repetition or a sound prolongation were lower than those of fluent control words, but these variables did not differ between stuttered and control words for single-syllable word repetitions.

2) **Howell (2007). People who recover increase the relative number of whole-word repetitions (singly or as parts of phrases – called *stallings*). People who persist do not increase the relative number of whole-word repetitions but do increase part-word repetitions, prolongations and broken words (called *advancings*). The higher rate of whole-word repetitions in recovered cases and the lack of change in persistent speakers suggests whole-word repetitions may have a role in promoting fluent speech**

3) **Howell, Bailey and Kothari (2010) (next slide).** Stalling and advancing symptom classes were examined separately in one-syllable function words (F1), one syllable content words (C1) and two syllable content words (C2). Percentage of the stalling and advancing classes were plotted across ages separately for persistent and recovered speakers Figure in next slide shows F1, C1 and C2 (top-bottom) with age on the abscissa and percentage of stutterings in each class on the ordinate (stallings and advancings and speaker group can be identified by the symbols given in the caption).

Slide 4

Evidence whether to include whole-word repetitions

4) **Manipulating whole-word repetitions by operant procedures impacts on fluency.** Howell, Au-Yeung, Charles, Davis, Thomas, Reed, Sackin and Williams (2001) and Reed, Howell, Davis, and Osborne (2007) used operant techniques to manipulate the incidence of stalling class disfluencies in children who stuttered. Increasing the relative proportion of stallings by the operant techniques reduced advancings.

5) Everyone uses Riley's (1994/2009) SSI-3/SSI-4 and the instructions for estimating severity specifically recommend whole-word repetitions are not considered as stutters

6) Yairi and Ambrose (2005) altered their assessment method that gave equal weighting to whole-word repetitions to that which other stuttered disfluencies to a weighted SLD which gives whole-word repetitions less of a role than other stuttered disfluencies by weighting them according to the number of repetitions they contain

Slide 5

Slide 6
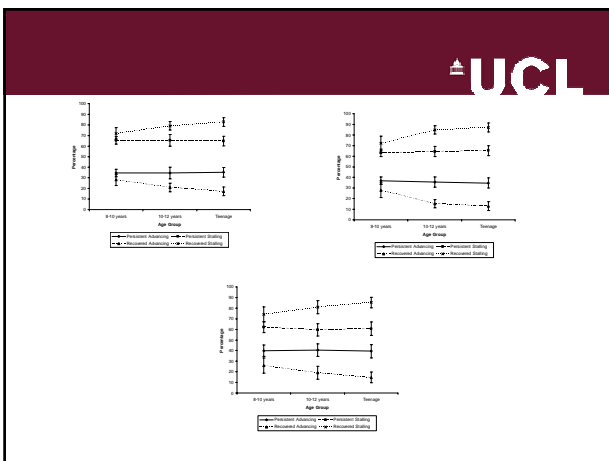
Issue 2 Symptom threshold

- 3-4% criterion
- 1) Yairi and Ambrose (2005)
- 2) Reilly et al. (2009)
- 3) Boey et al. (2009)
- The procedures used for assessing symptoms will have different sensitivity and reliability. Boey did not base on recordings, the other two did but there are sparse details about what procedures were used.
- Study that systematically compared different methods of assessing Riley's stuttered events (not SLD): Howell, Soukup-Ascencao, Davis & Rusbridge (submitted). Digital method more sensitive (more stutterings detected) than live method. Regression equation given so can convert live-digital procedure.
- The important point is that 3% means different things depending what procedure is used.

Slide 1

Famous people who stutter

Slide 2

Point is that disfluencies at 3% rate is not a lot as this a highly fluent individual shows

Slide 3

Two schemes that have:

1) inclusion criteria for initial diagnosis of stuttering (the state of stuttering pertinent to this topic)
2) assessment for persistence and recovery at the end of the study (next topic on change of state).

Examine different age ranges:

1) Yairi and Ambrose onset to about age eight years
2) Howell's team from age eight to teenage (plus)

Observations:

1) Complement each other with respect to age range
2) Together cover range onset to age past which recovery is unlikely (Andrews & Harris, 1964)

Warning:

Very different procedures adopted

Slide 4

Yairi and Ambrose's (2005) inclusion criteria

- The children in Yairi and Ambrose's work are obtained by parental referral and they tend to come from high socio-economic classes (Cook personal communication). A diagnosis of stuttering was given for children who met *all* the following criteria (Yairi & Ambrose, 2005, p.29):
  1. The child was six years of age or younger.
  2. Parents reported that they considered their child stuttered. The report was in the form of a parent rating of stuttering severity which had to be at least 2 on an 8-point scale (where 0 = normal, 7 = very severe).
  3. Yairi and Ambrose both considered that the child was stuttering.
  4. The child was given a severity rating of greater than one on the Illinois Clinician Stuttering Severity Scale (ICSSS) by the parent and the clinicians. The ICSSS and the way it is scored are described below.
  5. The child had to exhibit less than 3% of stuttering-like disfluencies (SLD). SLD consist of the following symptoms: Part-word repetitions, monosyllabic whole word repetitions and "disrhythmic phonation". The latter includes sound prolongations, blocks (audible or inaudible sound prolongation within words) and word breaks (Yairi & Ambrose, 2005, p.97).
  6. There was no reported history of neurological disorder.

Slide 5

Comments

- 1) Criteria 1 and 6 are objective.
- The cutoff age of six, used in criterion 1, is too low to include all children who stutter as onset of stuttering occurs above that age (Andrews & Harris, 1964).
- Using the low cutoff biases statistics – e.g. the age of stuttering onset would be estimated lower than it actually is.
- Criteria 2 and 3 are subjective and it is unclear how they were judged.
- SLD is vital to two of the diagnostic criteria (4 and 5):
- Criterion 4 - SLD are used as part of the ICSSS scale
- Criterion 5 - 3% SLD (no percentage is specified in criterion 4)
- For both criteria 4 and 5, the earlier observations about SLD apply.
- The 3% threshold seems low
- SLD includes whole-word repetitions that may be problematic symptoms.
- 2) The ICSSS criterion is examined in more detail next as it is critical to a diagnosis of stuttering.
- ICSSS gives a severity rating on an 8-point scale.
- This is obtained from:
- 1) an assessment of frequency of stuttering;
- 2) duration or repetition units;
- 3) rated tension of disfluencies;
- 4) secondary characteristics.

Slide 6

Comments Continued

- Frequency, duration and tension each receive rating values between 0 and 6.
- These are averaged to give a composite score which will also range between the values 0 and 6.
- The secondary characteristic score is then added in to give a final rating between 0 and 7 (an eight-point scale).
- The secondary characteristics weight less in the final severity score compared to the other three characteristics.
- There are numerous ways that a child could receive a score of greater than one that indicates stuttering:
- e.g. a child could have three whole-word repetitions in a 100 syllable sample which could receive a score of 1
- there would be insufficient SLD to obtain a duration score
- tension could be "slight" (this receives a score of 2).
- These have an average of one.
- The child might show no accessory characteristic, so the minimum score of 0.25 is given.
- Adding the 0.25 to the average score of 1 for the first three characteristics indicates that this child meets this criterion for stuttering.

Comments Continued

- There has been limited statistical assessment of the ICSSS (ten video tapes were assessed separately by Yairi and by Ambrose), which indicates good reliability (no assessments of internal and external validity have been made and the scale is not standardized).

- 3) Generally speaking the criteria employed suggest that children who do not stutter may be designated as stuttering. It is a cautious criterion (inclusion of all suspected cases as people who stutter). There is a potential ethical problem insofar as lenient criteria may lead to children being designated as stutterers who are fluent. This could possibly have a traumatic effect on the child's subsequent development.

Howell (in press)

- Howell's group works with clinics that receive referrals from general practitioners, other speech-language pathologists and self-referrals. The diagnoses are based on judgments by clinicians as follows:
- 1) Children were aged between 8 and 10 years
- 2) An initial diagnosis of stuttering was made by a trained speech pathologist who worked independent of the clinicians who examined the child subsequently
- 3) The child was referred to a clinic that specialized in childhood stuttering
- 4) The specialist clinic confirmed the diagnosis and admitted the child to group (rather than individual) treatment
- 5) There was no reported history of neurological disorder.

Comments

1) Criteria 3 and 4 require care in judgment insofar as they entail putting cash resources into the child's treatment at each of these levels

2) Treatment and follow-up were kept constant for all participants

3) The child was recorded before any treatment started so the initial recordings were not affected by treatment.

4) Speech symptomatology was deliberately excluded as a diagnostic criterion at inclusion so that it can be examined as a risk factor for persistence/recovery.

Topic two Passing out of the state of stuttering or not at later ages

Yairi and Ambrose (2005)
Children who recover

Yairi and Ambrose designate a child as recovered if he or she met *all* of the following criteria after a minimum period of four years since onset of the disorder:

1. Parental report that the child did not exhibit stuttering.
2. Parental rating of zero (normally fluent) on the "rating scale described in chapter two" (p.164). Chapter two describes the ICSSS, but this is unlikely to be the scale referred to as it appears to be specifically for clinicians' use. The only other mention about the parent rating scale in chapter two (p.35) directs the reader to chapter three. Chapter three just mentions the end-points of the scale on page 71 (there is further comment on p.74, but this does not shed any more light on the parent rating scale).
3. Clinician's general judgment that the child was not stuttering.
4. A severity rating of zero on the ICSSS by a clinician
5. Fewer than 3% SLD
6. No remission of stuttering for 12 months after this assessment for persistence/recovery as judged by parent and clinician.

Comments:

- 1) The four-year follow-up period should have been specified as an inclusion criterion for the initial phase of the study. Without this, it is not possible to obtain the following important pieces of information: 1) Attrition rate; 2) Selective attrition (whether recovered speakers were selectively lost); 3) Recovery rate over the period from initial inclusion to the four-year follow-up assessment.
- 2) The points made about symptom assessment earlier apply here too: 1) 3% is a low threshold requirement; 2) The doubtful/problematic whole-word repetitions are included; 3) It is not clear how the symptom assessments were made. The chance of children being classified as recovered is minimized in the way symptoms are assessed. Ethical issues about whether it is appropriate to designate someone as stuttering when they do not suffer from the disorder arise again.
- 3) Use of low ("borderline") rating scale criteria for parent rating also minimizes the chance of misdiagnosing a speaker as recovered.
- 4) Criteria 4 and 5 should not be used if speech symptoms are to be used as a risk factor for recovery from stuttering (otherwise the reports are circular).

Comments Continued

- 5) Criterion 5 is not independent of criterion 4.
- 6) Investigations which do not start from stuttering onset cannot produce an absolute estimate of recovery rate (Yairi & Ambrose, 2005).
- 7) Investigations which do not extend to teenage when developmental stuttering is resolved (Andrews & Harris, 1964) cannot provide definite estimates of recovery rate. As noted, the Yairi and Ambrose study does not require speakers to be followed up to a particular age, so such estimates are not provided. Based on modal age of onset and the requirements that the children continue in the study for four years after onset of the disorder, it would appear that testing ceases at about age 7-8 years. Onset of stuttering occurs in some children after that age (Andrews & Harris, 1964) and recovery rate in the period 8-teenage is approximately 50%. Cases of late onset who subsequently recover and cases who recover between 8 and teenage will be missed.
- 8) Most of the children received treatment and the type of treatment varied between children (Yairi & Ambrose, p.36). Consequently, the impact of treatment on recovery cannot be determined (**it is unlikely that the recovery is "natural"**)

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Children who persist

- A child persisted if he or she met *any* of the following criteria after a minimum period of four years since:
  1. Parent report of stuttering episodes
  2. Parent rating of higher than one
  3. Investigators' observation of speech characteristics that they judged as stuttering
  4. A severity rating of higher than one on the ICSSS

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Comments

- 1) Here the inclusion of whole-word repetitions maximizes the chance of someone being called persistent.
- 2) Use of low rating scale criteria maximizes the chance of persistent stuttering.
- 4) The requirement to meet one criterion alone allows persistence to be based solely on parental report).
- 5) SLD is used implicitly in ICSSS but not explicitly elsewhere.
- 6) It is not possible to designate a child with a score of 1 as persistent or recovered.

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Main findings

- They comment: "The ability to make accurate predictions could have a revolutionary impact on the long-term objective of cost-effective selective treatment for stuttering children. It is not practical, possible, or necessary to put every child who stutters into therapy. Economic conditions and emerging health policies, in fact, may make this option more difficult. For any child who appears likely to continue to stutter treatment should not be delayed. But it may be advantageous to defer treatment for children with few or no risk factors and/or mild stuttering that does not cause concern for either child or parents." (Yairi, Ambrose, Paden & Throneburg, 1996, p. 74).

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Comments

1. Risk factors are not 100% accurate. Parents and clinicians may make the wrong decision based on this information. Some clinicians have represented the view that they would prefer not to have such information rather than face this ethical dilemma
2. Procedural issues may also suggest that some children are included in the study who do not stutter (use of whole-word repetitions). potentially misleading to clinicians that might again raise ethical concerns.
3. Procedural factors that bias against classifying speakers as recovered as indicated above and towards calling speakers persistent (e.g. use of low rating scale criteria for parents)

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Howell (in press)
Children who recover
- Based on questionnaires. Developed, assessed and standardized parent and child questionnaires and a researcher assessment form that mimicked what therapists indicated they take into account when assessing whether a child stutters or not on more than 300 participants. Construct reliability, test-retest reliability, internal validity and external validity (assessed against SSI-3) have been measured and standardized scores have been reported the questionnaires and external validity and standardized scores for the researcher assessment (Howell et al. 2008b).
- A child was designated recovered if he or she met *all* of the following criteria at teenage:
  1. Child indicated he or she was not stuttering on the child report questionnaire
  2. Caregiver indicated that the child was not stuttering on the parent report questionnaire
  3. Researcher indicated that the child was not stuttering on the researcher assessment form
  4. Neither the child, caregiver nor researcher reported any remission for at least a year after reaching teenage

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Comments

1. Status of whole-word repetitions is avoided (they can be examined empirically).
2. SSI-3 used for external validation. Can see what SSI-3 scores are equivalent to questionnaire/ researcher assessment. 1) Caution that appear to show stuttering because Riley doesn't have a no-stuttering class; 2) Also digital are sensitive means get higher scores than with other procedures; 3) Have assessed fluent speakers using our procedures and recovered not statistically different to them (Davis, Shisca & Howell, 2007). External validation that stuttering is like fluent speech.

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Children who persist

- A child persisted if he or she met *all* at teenage:
- Child indicated stuttering on the child report questionnaire
- Caregiver indicated t stuttering on the parent report questionnaire
- Researcher indicated stuttering on the researcher assessment form
- Neither the child, caregiver nor researcher reported any recovery for at least a year after reaching teenage

Comments

- 1) Have to meet al criteria either for recovery or persistence.
- 2) Symptom type, frequency of occurrence etc. can be examined as risk factors (not circular).

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Main findings

- 1) 50% recover in this age range.
- 2) Howell, Bailey and Kothari (2010) looked at the way the proportion of stalling and advancing changed over three ages for persistent and recovered speakers.
- These data offer information about two questions. 1) The first session data can be examined to see whether the children at risk of persisting could be distinguished from those who will recover. NB pre-intervention; 2) Trends in the data across sessions indicate the subsequent course of the disorder.
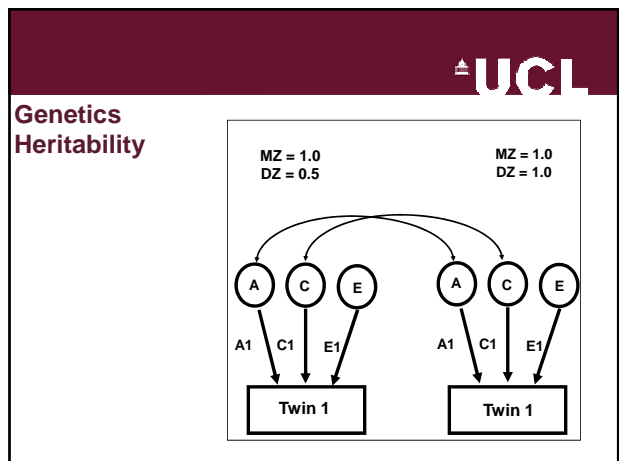

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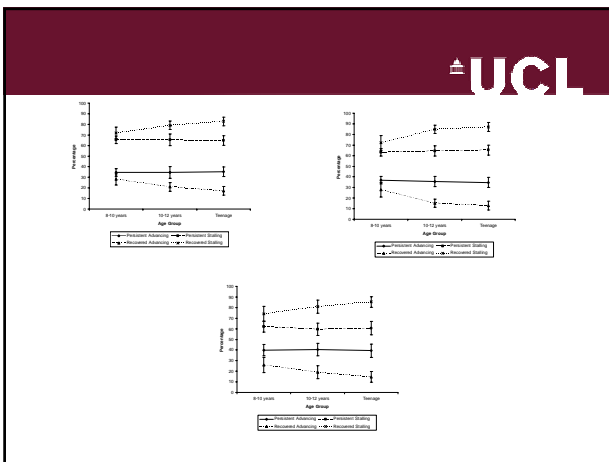
1) High rates of stalling are associated with subsequent recovery. Also means that high rates of word repetition are also associated with recovery. The ratio of stallings to advancings or whole-word repetitions to part-word repetitions, prolongations and broken words appears to be an important risk factor for subsequent recovery.

2) Across ages the recovered speakers have an increased proportion of stallings. The proportions did not change for the persistent children. Gradual changes occur in speakers who recovered from their stuttering. Howell et al. (2001) suggested that whole-word repetitions may have a role in facilitating fluency

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Topic, transition between stuttering and fluent states

- How speakers get from one state (stuttering) to the other (recover) or not (persist). The Howell et al. (2010) study just discussed already showed that speakers who recover show a gradual change in disfluency type.
- Wingate maintained that:
    1) "recovered" cases were misdiagnosed because the wrong symptoms were used (whole-word repetitions). As these resolved, this appeared to suggest incorrectly that recovery had occurred;
        2) though the disorder cannot be cured it can be managed (allowing symptoms to ameliorate possibly temporarily).
- **Persistent stuttering – factors that might support the view that change is not possible**
        Arguments that there is something organically different about speakers who persist in their stuttering has come from genetics, brain imaging and drugs work.

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Genetics Heritability

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- **Findings:**
- All studies suggest 70/30 split heredity/environment (Andrews, Morris-Yates, Howie, & Martin, 1991; Dworzynski et al., 2007; Felsenfeld, Kirk, Zhu, Statham, Neale & Martin, 2000; Ooki, 2005).
- **Linkage analyses:**
- Alleles from related family members are identical by descent (IBD) if they are copies of the same allele that was passed on from the same ancestor.
- Both siblings show the trait in question, IBD alleles that the siblings share are candidates for harboring the mutant form of the gene responsible for that trait.
- The observed IBD pattern of affected siblings is compared to a baseline pattern predicted from random linkages, stretches of DNA can be located that are co-inherited by affected individuals at above chance level.
- Significant linkage has been reported on chromosome 12 in a study of stuttering in 46 consanguineous families from Pakistan (Riaz et al., 2005).

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Association analyses:
- Allelic frequencies are compared for groups such as individuals with the disorder versus controls or low-scoring versus high-scoring individuals with respect to a quantitative trait
- The Pakistani family PKST72 used by Riaz et al, the linkage to chromosome 12 was first replicated with additional family members included.
- Association analysis was then carried out on the linkage region of the long arm of chromosome 12 (Kang et al., 2010).
- The study scanned 45 genes in several affected individuals.
- They identified a missense mutation, i.e. a single base pair substitution that changed the code for one amino acid into another, in the *GNPTAB* gene. They went on to screen for this and other mutations in the *GNPTAB* gene in unrelated probands in Pakistan.
- They also screened the same region in 270 unrelated individuals affected by stuttering from North America and Britain and 276 unaffected North American subjects as controls.
- The authors identified additional mutations in the *GNPTG* and in the *NAGPA* genes. In all cases, the three mutant genes occurred at significantly higher rates in the stuttering samples than in the non-stuttering control samples.
- The genes that were identified are involved in lysosome processes in cells. The lysosome contains enzymes which help to recycle substances, such as food particles or engulfed viruses etc, into material that the cell can utilize.
- In the case of the study on stuttering, it is emphasized that there were people in the sample who carried the mutation and were not affected by stuttering (incomplete penetrance) and there were also individuals who stuttered without any of these mutations (phenocopy).

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Next Stages

- Need to trace the function of the gene back to brain circuitry as has been done in the famous case of the KE family who have another speech disorder (not stuttering). The speech problems in this family are associated with a mutation on the *FOXP2* gene. When DNA binds to *FOXP2* the function of certain target genes can be activated or repressed. A protein that can switch other genes on or off in this way is called a transcription factor.
- The functions of *FOXP2* have been traced back to brain circuitry and the impact this has on behavior has been established using animal models where certain genes are 'knocked out' (mice have pup vocalization problems Groszer et al., 2008) zebra finches that have decreased *FOXP2* functioning (they had difficulty learing new tunes Haesler et al., 2007) and cans on the KE family..
- Debate about whether this suggests that early intervention is useful or not. Analogy with the work on another genetic disorder, tuberous sclerosis complex (TSC), suggests that behavioral interventions may be effective in diseases where there is a known genetic influence. TSC can be treated with the drug rapomycin and that the drug operates on mechanisms that are downstream from the genes themselves (de Vries & Howe, 2007; Napolioni, Moavero & Curatolo, 2009).

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Behavioral

- Consider this before scanning as need some of it to understand scanning studies I will discuss
- The Howell et al. (20010) data show:
  1) that recovered speakers make a change whereas persistent do not;
  2) that there were differences that were not a response to the treatment (the first recording);
  3) the difference between speaker groups in terms of symptoms was not due to circularity because they were used at initial diagnosis and during subsequent assessments;
  4) they were obtained by properly assessed instruments.
- The only possibility that I wont have time to discuss is that the participants were selected either by self or by therapists as based on their assessment of their chance of recovery (Lieckfeldt**).**

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Questions:

1. What could the role of whole-word repetition be?
2. Why might this facilitate fluency in the recovered group?
3. Why do persistent speakers not do what the recovered speakers do?
4. Other established facts that theory needs to be consistent with are experimental interventions that induce fluency (such as FAF that I discovered).

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- Language planning and speech motor programming and execution are both widely regarded as being affected in people who stutter. Usually either planning or execution processes are considered as being involved. However recent models include both processes (Howell, 2004; Howell, 2007; Howell and Au-Yeung, 2002). Lu, Chen, Ning, Ding, Guo, Peng, Yang, Li and Lin's (2010) work follows this approach and extends it by examining the neural substrates of planning and execution. According to Howell's theory, some stuttering symptoms are related to the planning processes and some are associated with execution processes as indicated by the different roles each symptom class plays with respect to dealing with dysfluency. In particular, whole-word repetitions (and also pauses) are ways of stalling motorically (repeatedly executing a previously generated program) on material prior to other material that is difficult to plan) whereas prolongations part-word repetitions and word breaks reflect planning problems (the repetition, prolongation and hesitation within words signify that the plan was not right or was only partly prepared). Prolongations, part-word repetitions and word breaks are referred to as advancing to indicate the speaker has moved forward prematurely in the speech stream and to contrast with what happens in stalling. The theory is called EXPLAN to acknowledge the contributions of motor execution and language planning. Unlike the perspectives that included or excluded whole-word repetitions as characteristics of stuttering, this perspective gives whole-word repetitions a different role (motor based) to symptoms that reflect planning problems. Although this theory focuses on language and motor processes, it should be noted that stuttering affects other things too (e.g. socio-emotional and non-speech cognitive behavior).

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- Part of the motivation for developing EXPLAN theory was the fact that neither language nor motor processes alone can exclusively account for why stuttering starts in children and why it persists in children who are still stuttering at teenage. No milestones in language development have been identified that explain why stuttering starts (Howell, in press a). For instance, the average age of stuttering onset is about three years of age (Yairi and Ambrose, 2005), by which time most syntactic development is complete (Limber, 1973). The subsequent course of stuttering shows no marked problem in planning. For instance, children who continue to stutter do not exhibit dysarthria, oral dyspraxia, or other signs of an incorrect mental model of the utterance (Brown et al., 2005). The speakers who persist do not show any major abnormality of the speech motor system (motor cortex, basal ganglia, lower motor neurons), as there is no oral weakness, spasticity, tremor, or hypophonia (Brown et al., 2005).

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- The distinction between language and motor levels in EXPLAN allowed planning (e.g. language processing deficits) and/or execution (motor timing deficits) problems to result in stuttering. Planning and execution have to link together (interface) for meaningful speech to be output. According to EXPLAN, the interface accounts for how failures of planning or execution can result in stuttering. The way the model operates is illustrated with the utterance "I split it". The speaker starts by planning "I" which is a simple word. "I" is a word from the function word class and words from this class are, generally speaking, easy to plan so they are generated rapidly. Once the plan is ready, it can be sent to the motor execution processes where the motor programme can also be generated rapidly after which this word can be output. Planning and execution are independent in the model, so whilst motor preparation for this word is taking place, planning for the verb "split" can be performed. "Split" would be more difficult to plan than "I". "Split" is a content word and these types of words are, generally speaking, harder to plan and take more time to prepare than function words. In this particular case, the difficulty arises because the verb has a consonant cluster and starts with the phone /s/ that is amongst the latest sounds that a child learns. Since "I" can be output rapidly whilst planning for "spilt" can take some time, the plan for the latter may not be ready at the time that execution of "I" is completed. When this occurs, the plan for "split" is incomplete after "I" has been uttered.

⛫UCL

- **Findings** All studies suggest 70/30 split heredity/environment (Andrews, Morris-Yates, Howie, & Martin, 1991; Dworzynski et al., 2007; Felsenfeld, Kirk, Zhu, Statham, Neale & Martin, 2000; Ooki, 2005).
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- The study scanned 45 genes in several affected individuals.
- They identified a missense mutation, i.e. a single base pair substitution that changed the code for one amino acid into another, in the *GNPTAB* gene.
- They went on to screen for this and other mutations in the *GNPTAB* gene in unrelated probands in Pakistan.
- They also screened the same region in 270 unrelated individuals affected by stuttering from North America and Britain and 276 unaffected North American subjects as controls.
- The authors identified additional mutations in the *GNPTG* and in the *NAGPA* genes. In all cases, the three mutant genes occurred at significantly higher rates in the stuttering samples than in the non-stuttering control samples.
- The genes that were identified are involved in lysosome processes in cells. The lysosome contains enzymes which help to recycle substances, such as food particles or engulfed viruses etc, into material that the cell can utilize.
- In the case of the study on stuttering, it is emphasized that there were people in the sample who carried the mutation and were not affected by stuttering (incomplete penetrance) and there were also individuals who stuttered without any of these mutations (phenocopy).

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Next stages
Need to trace the function of the gene back to brain circuitry as has been done in the famous case of the KE family who have another speech disorder (not stuttering). The speech problems in this family are associated with a mutation on the *FOXP2* gene. When DNA binds to *FOXP2* the function of certain target genes can be activated or repressed. A protein that can switch other genes on or off in this way is called a transcription factor. The functions of *FOXP2* have been traced back to brain circuitry and the impact this has on behavior has been established using:
1) animal models where certain genes are 'knocked out' (mice have pup vocalization problems Groszer et al., 2008)
2) zebra finches that have decreased *FOXP2* functioning (they had difficulty learning new tunes Haesler et al., 2007)
3) scans on the KE family..
Debate about whether this suggests that early intervention is useful or not. Analogy with the work on another genetic disorder, tuberous sclerosis complex (TSC), suggests that behavioral interventions may be effective in diseases where there is a known genetic influence.
TSC can be treated with the drug rapomycin and that the drug operates on mechanisms that are downstream from the genes themselves (de Vries & Howe, 2007; Napolioni, Moavero & Curatolo, 2009).

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Behavioral
- Consider this before scanning as need some of it to understand scanning studies I will discuss
- The Howell et al. (20010) data show:
- 1) that recovered speakers make a change whereas persistent speakers do not;
- 2) that there were differences that were not a response to the treatment (the first recording);
- 3) the difference between speaker groups in terms of symptoms was not due to circularity arising because they were used at initial diagnosis and during subsequent assessments;
- 4) they were obtained by properly assessed instruments.
- The only possibility that I wont have time to discuss is that the participants were selected either by self or by therapists as based on their assessment of their chance of recovery (Lieckfeldt**).**

Questions:
- What could the role of whole-word repetition be if they are not typical stutters?
- Why might this facilitate fluency in the recovered group?
- Why do persistent speakers not do what the recovered speakers do?
- Other established facts that theory needs to be consistent with are experimental interventions that induce fluency (such as FAF whose fluency-enhancing effects were *first reported* by Howell, El-Yanin and Powell, 1987).

EXPLAN theory
- In general, language planning and speech motor programming and execution are both widely regarded as being affected in people who stutter.
- Usually either planning or execution processes are addressed by speech-language therapists. One of these alone does not seem sufficient to account for stuttering although both possibly have a role
- Language – content words are more problematic for persistent stutterers than are function words
- Motor – problems of timing and coordination

1. EXPLAN incorporate language planning and execution and the interface.

2. EXPLAN incorporate language planning and execution and the interface.

3. EXPLAN incorporate language planning and execution and the interface.

- Illustrate way EXPLAN operates with "I split it" (FCF).
- The speaker starts by planning "I" which is a simple word.
- "I" is easy to plan and once ready, it can be sent to the motor execution
- The motor programme can also be generated rapidly after which this word can be output.
- Planning and execution are independent in the model, so planning for the verb "split" can be performed while I is being output.
- "Split" is a content word and takes more time to prepare.
- The plan for "split" may not be ready at the time that execution of "I" is completed.
- The execution processes can be stopped (paused) or the speaker can repeat the current plans of words that were just received (this results in whole-word or phrase repetition).
- Children who stutter, like fluent children, begin by stalling as just described when they encounter problems generating difficult items like "spilt".
- If, however, the stuttering persists, symptom types change: Speakers then start to produce disfluencies on content words and these are different from stallings. Examples often seen in persistent developmental stuttering are "I sssplit it", "I s.s.split it" and "I s/plit it" (where / indicates a break within the word.
- According to EXPLAN, these all occur because these speakers have stopped stalling and attempt the word on the basis of an incomplete plan.

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Neural substrate
- The neural mechanism responsible for execution is the cerebellum.
- The cerebellum controls speech timing.
- Children who stutter show performance deficits in the Wing-Kristofferson (1973) isochronous tapping task that work in patient with cerebellar damage has linked to the cerebellum (Ivry, 1997).
- Work has also shown that children who stutter perform worse than controls on the Dow-Moruzzi battery of tasks that assess cerebellar performance (Howell, Davis and Williams, 2008).
- Unusual cerebellar activity is always observed in scans of people who stutter.
- Delaying voice feedback disrupts cerebellar processes (Howell and Sackin, 2002) and this may explain the positive effects of this manipulation on the fluency of people who stutter.
- EXPLAN only specified the substrate for execution and considered abnormal activity in this structure in people who stutter is functional (later we'll see Lu take these ideas further).

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- **How execution receives inputs, programmes and executes them and checks that the plan for successive utterances was a complete one, how this changes over development and why this is reflected in changes in symptom type (stallings to advancings).**
- An efferent copy of any speech issued for output is taken (Howell, 2002).
- It is subtracted from the plan after execution is completed.
- If the speaker started with the plan for a whole utterance, this was issued and there would be no difference.
- There would be a difference if the speaker started with an incomplete plan (as happens during advancing) because the incomplete plan is updated during the time for execution.
- The difference is not cancelled so a signal is generated that alerts the speaker that incomplete plans are being received and adjustment to speech timing is required.

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- This process would work in a person who persists in their stuttering as well as in fluent speakers during early development
- Why do persistent speakers become less sensitive to the alerts?
- Answer: physiological adaptation (Howell et al., 2000): By definition, speakers who continue to stutter would produce a lot of disfluencies that in turn lead to many alerts. Speakers who persist would ignore them, more would be produced, they would be adapted to and gradually the speakers who persist become less sensitive to the alerts. De facto, the alerting process ceases working

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Priming function and content words.
- Priming allows an experimenter to manipulate planning time by getting them to repeat something similar to what they did previously. Savage and Howell (2008) priming of utterance like 'He is swimming' (FFC).
- When the auditory prime matched an aspect of the probe (e.g. 'he is' was primed and the picture was of a boy, or 'swimming' was primed and this was the action), the planning time needed for the production of the primed elements in the phrase was reduced.
- Predictions:
- 1) Priming a function word reduced its planning time, allowing it to be produced more rapidly. When rate of production of a function word is increased, pressure is placed on having the content word plan ready earlier. The reduction of planning time for the function word would enhance the time pressure for preparing the content word, and increase the chance that it is produced disfluent;y (of either the stalling or advancing type).
- 2) Priming the content word reduces its planning time (as with function words). But this time priming should reduce the disfluency rate on function and content words (priming the content word accelerates planning and decreases the chances of plan-unavailability, which should be reflected in a reduction of disfluency on the function or content word).
- Savage and Howell (2008) confirmed these predictions in children who stuttered and controls (mean age six years). Priming function words increased disfluencies on function and content words whereas priming content words reduced disfluencies on function and content words.

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- **Scanning**
- Lu, Chen, Ning, Ding, Guo, Peng, Yang, Li and Lin's (2010) neural substrates of planning, execution and the interface:
- Task variable associated with planning - "number of syllable sized motor programs".
- Task variable associated with execution was "syllable size and onset complexity".
- fMRI scans were taken as the task was performed.
- Comparison was made between the participant groups to determine what were the neural substrates of planning, execution and the interface.
- The authors used the task-by-speaker group interactions to identify aberrant planning or execution processes in people who stutter.
- Structural equation modeling was used to identify the neural substrates that were associated with each of the processes.
- Structures that were associated with planning were the bilateral inferior frontal gyrus and the right putamen
- Structures that were associated with execution were the left premotor area and anterior gyrus, right insula, and the cerebellum.
- The structures that were associated with the interface were the left premotor area and anterior gyrus and the right insula.
- **Computational modeling**
- Bayesian implementation of EXPLAN – 100 % accuracy with training data and 85.7% with test data (Ravikumar, 2009).

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Practical issues brief comments
- 1) No closer to distinguishing natural and treatment-assisted recovery. Norbert Lieckfeldt has expressed the opinion that there is some selectivity in which children seek treatment and which children therapist seek to treat with those less likely to recover to be more prone to be seen in clinic in each case. There is no evidence to support this.
- 2) Many people are arguing for subtyping. Looking for someone with joint disorders (comorbidity that lies behind many proposed subtypes) is going to make location of clearcut cases for study more problematic. For example, looking for someone who stutters and who has ADHD and, say, each can be reliably identified 75% of the time would lead to joint cases being reliably identified 56% of the time. Psychologists are moving away from comorbidity and looking at networking approaches (how local connections between symptoms build up). There are potentially some interesting implications of this for stuttering (Howell, 2010).

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Summary and conclusions
- **State**
  - Empirical work suggests that whole-word repetitions have a different role to more typical disfluencies
  - 3% stuttering seems a low criterion
  - Schemes that addressed inclusion and end state were examined (Yairi and Ambrose, 2005; Howell, 2010). The procedures employed are very different.
  - Some of the main issues – avoid circularity when assessing risk factors, define stuttering and outcome so they are not biased either to recovered or persistent forms.
- **Change state**
  - Symptom type as risk factor that predicts outcome at teenage
  - Possible ethical issues associated with state definition and with respect to whether clinicians want to be able to predict outcome
- **Process**
  - Consider that recovery occurs (people were in the state and gradual changes in symptom type have been documented
  - Wingate's alternative viewpoint – misdiagnosis and why recovery can appear to happen
- Genetic, behavioural and scanning assessments of some of the processes involved in stuttering recovery and work that needs to be done
  - Genetics – heritability pretty much settled, linkage and association results suggestive, but incomplete penetrance and phenocopy are issues
  - Main thing is that need similar work to what has been done with the KE family (knock-out, decreasing gene function and scans of affected human cases).
- Behavioural – formal model that been tested and supported in various ways: covered priming, Lu and Ding's scanning work and Indian work on automatic recognition of stuttering.
- Briefly mentioned problems and directions forward with respect to some practical issues (subtyping, networking and the natural-treatment assisted recovery distinction).

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References

Students who wish to read more about the ideas I presented can follow up the ideas in the following publications:

Howell, P. (2004). Assessment of some contemporary theories of stuttering that apply to spontaneous speech. *Contemporary Issues in Communicative Sciences and Disorders*, *31*, 122-139.

Howell, P. (2010). Language processing in fluency disorders. In J. Guendouzi, F. Loncke and M. Williams (Eds.). *The handbook on psycholinguistics and cognitive processes: Perspectives on Communication Disorders* (pp.437-464). London: Taylor & Francis.

And my book (out October 2010):

Howell, P. (in press). *Recovery from Stuttering.* Psychology Press. 978-1-84872-916-2.

Other publications are on my group's website:
http://www.speech.psychol.ucl.ac.uk/