

Cluttering and the mentally challenged

Key-note

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How it started?

How it started?

- Reference is made to stuttering - as one of the primary characteristics in the description of various syndromic intellectual disabilities.
- Specifically, Down syndrome and Fragile-X syndrome include "stuttering" as a characteristic of speech. In recent years this understanding has come under increasing scrutiny by researchers and clinicians.

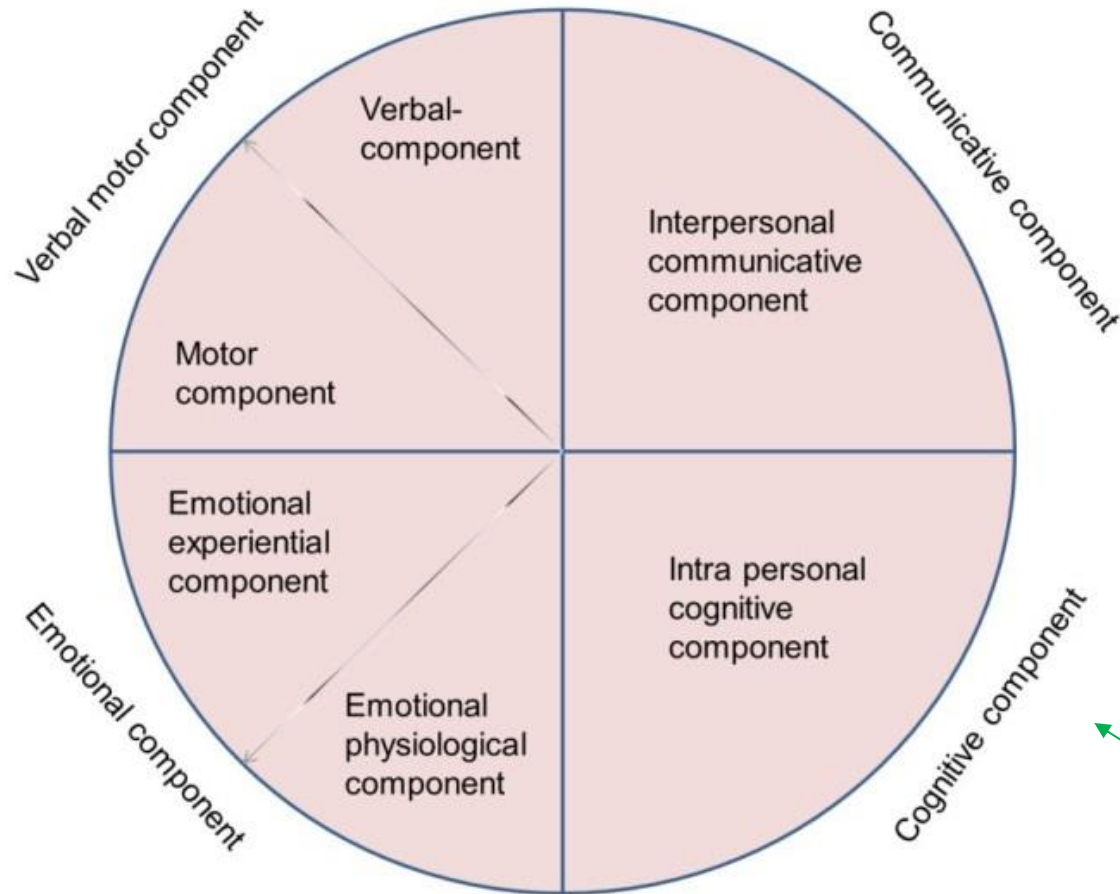
What is in the name?

- Mentally retarded
- Mentally challenged
- People with intellectual disability

What components have to be considered in fluency and disfluency assessment?

Four components

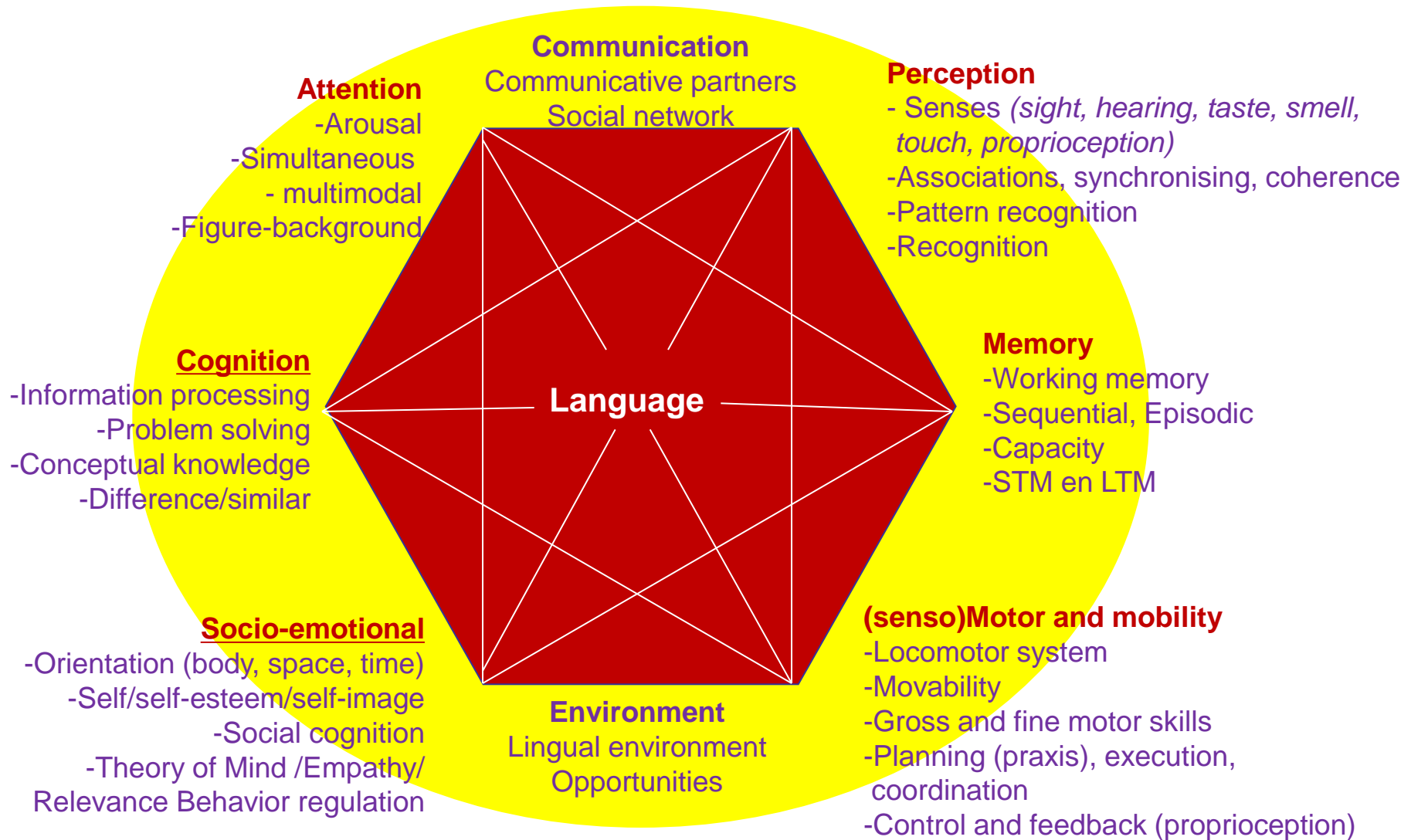
(Stourneras, 1979)



IQ
??

- Influences on communicative competency

Domains of Communicative Competence



Stuttering (Starkweather, 1987)

1 an abnormally high frequency and
abnormally long durations of sound,
syllable and word repetitions

or

2 an abnormally high frequency and
abnormally long durations of
prolongations & pauses

Cluttering

- A fluency disorder in which a person is not able to adjust rate to the language or motor demands of the moment (van Zaalen, 2009)
- Symptoms: fast an/or irregular rate; NDF, errors in pausing or unintelligibility (St. Louis et al., 2010)

Subtyping (Ward, 2006; van Zaalen, 2009)

- Syntactical Cluttering
- Phonological Cluttering

Disfluency and Down Syndrome

Down syndrome (DS) results from a triplication of part of Chromosome 21



* www.radiodownsound.com

Craniofacial anatomy: a compact mid- and lower-face skeleton, macroglossia, and a palate that is high and often shelf like.

The developmental trajectory of orofacial characteristics is not well established => fluctuating dental asymmetry (Barden, 1980).

- Approximately 1/800 live births (Roizen, 2002).
- DS is the most common single cause of intellectual disabilities (ID) (Menkes & Falk, 2005), with between 70% and 75% of individuals IQ of between 25 and 50 by the first decade of life (Vicari, 2006).
- associated with a distinct profile of developmental outcomes regarding body functions and activity performance (Fidler, Hepburn, Mankin, & Rogers, 2005), with evidence for great variation in the range and level of deficits resulting from biological and environmental factors (Turner & Alborz, 2003).
- specific motor deficits that are thought to contribute to activity limitations, including balance, coordination, and fine manipulative skills (Wuang & Su, 2000)



Down Syndrome

Limitation	Skill
Auditory perception	Visual perception
Expressive language	Receptive language
Gross motor	Non verbal functioning

Speech intelligibility is reduced:

Fast variable movements in multisyllabic words is limited. Anatomy, hypotony in the mouth area, a phonological delay, dyspraxia, and/or hearing loss (Kumin, 2006).

Relative weaknesses on items with verbal direction that involve semantic content, general comprehension, social intelligence, and reasoning.

Auditory digit span abilities are weaker than those of children with mental retardation of other etiologies matched for general cognitive abilities (Jarrold & Baddeley, 1997; Mackenzie & Hulme, 1987; Marcell &

Weeks, 1988; Varnhagen, Das, & Varnhagen, 1987) ■

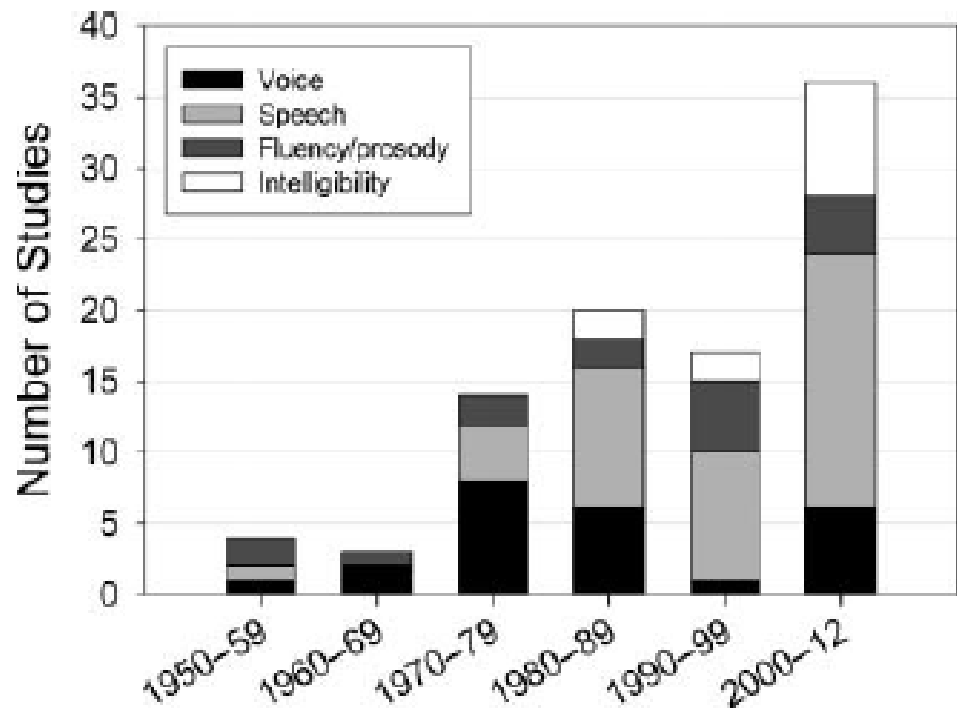
DS and Language domain

- Contrary to controls DS show more poorly on Number Recall and Word Order than they did on Gestalt Closure and Hand Movement, Pueschel, Gallagher, Zartler, and Pezzullo (1987)
- Language production does not keep pace with language comprehension, and syntactic abilities are an area of particular weakness. Language comprehension abilities, in contrast, continue to develop with age for individuals with Down syndrome. Chapman (1995)
- Contrary to controls, by adolescence, mean PPVT–R age equivalent was significantly higher than mean nonverbal mental age (Chapman, Schwartz, and Bird (1991) .

- DS and speech and disfluency

Speech Impairment in Down Syndrome: A Review

Kent et al (2012)



Disfluency and DS

Studies of speech disfluency in more than 300 participants have demonstrated that stuttering and/or cluttering occurs in individuals with **DS** at rates of **10%–45%** (Devenny&Silverman, 1990; Gottsleben, 1955; Keane, 1970; Preus, 1972; Rohovsky, 1965; Schlanger & Gottsleben, 1957), compared with the incidence of about 1% in the general population (Guitar, 1998).

It is generally not possible to distinguish normal developmental disfluencies from genuine stuttering or cluttering in this literature.

The published data do not permit conclusions on the persistence or developmental pattern of fluency disorders in individuals with DS.

Survey data confirm a rather high incidence of stuttering in individuals with DS: 17% in Kumin's (1994) parent report survey and 15.6% in Schieve et al.'s (2009) analysis of data from the National Health Interview Survey (Botman, Moore, Moriarity, & Parsons, 2000).

DS: <http://www.youtube.com/watch?v=RwIXyoHMfYA>

Rohovsky (1965) **observed a rate of 36% in individuals with DS who were institutionalized, compared with 19% in those individuals with DS who were not institutionalized.**

Similarities and differences in the types of disfluency

DS (n=19) **institutionalized**, compared to controls: disfluency patterns similar to beginning developmental stuttering Otto and Yairi (1974).

Willcox (1988): “it is clinically more appropriate to consider the speech non-fluencies of Down’s syndrome individuals as part of a global language deficit rather than as a symptom of the syndrome” (p. 169).

Stuttering or Cluttering in DS?

Cabanas (1954) *“the rhythm disorders in the individuals whom he studied should be called “cluttering” because of their restricted vocabularies, rapid speech patterns, and “lack of ideomotor equilibrium” (p. 36).*

Cluttering and DS

- Preus (1972) noted that both stuttering and cluttering occur in individuals with DS.
- Van Borsel and Vandermeulen (2008) classified a very large percentage of their 76 participants with DS as being either clutterers (about 80%) or clutterer–stutterers (about 17%).

Appendix C (p. 2 of 3). Summary of studies of fluency and prosody in individuals with DS.

Source	Participants	Method	Summary of results
Willcox (1988)	n = 5 DS* (3 M, 2 F; range = 10;10–15;01) n = 5 TD** (all M; range = 2;00–2;08) *Considered nonfluent. **Matched for language.	Perceptual ratings: Analysis of frequency and type of disfluencies	Similarities and differences observed in the disfluency types of the 2 groups. The mean number of nonfluencies for DS was 7.4 (per 100 words) and 3.6 for TD. Results were questionable because of individual differences. Repetitions were most common for both groups. Percentages of prolongations were much lower in the TD group than in the DS group.
Pettinato & Verhoeven (2008)	n = 16 DS (10 M, 6 F; range = 11.00–20.00) n = 12 TD* (range = 4.06–7.00 yrs) *Matched on receptive vocabulary level with sex balance similar to that for DS group.	Perceptual ratings: Examination of the production (using a nonword repetition task) and perception of word stress (using XAB discrimination task)	DS had processing difficulties in both the production and perception of more difficult and later acquired stress patterns as well as weak word-initial syllables.
Van Borsel & Vandemuelen (2008)	n = 76 DS (51 M, 24 F, 1 unknown) Range = 3.8–57.3 yrs M _{age} = 22.8 yrs	Perceptual ratings: Used the Predictive Cluttering Inventory (Daly, 2006), which was administered by 26 SLPs.	78.9% of DS had scores that classified them as clutterers, and 17.1% of DS had scores that classified them as clutterer-stutterers.
Gottleben (1955)	n = 36 DS (23 M, 13 F) Range = 8;11–51;07 M _{age} = 27;03 n = 36 AD* (23 M, 13 F) Range = 9;07–76;05 M _{age} = 28;03 *Individuals with mental retardation but not DS.	Perceptual ratings: Judgments of stuttering by 3 individuals	33% of DS were identified as stutterers.
Schlanger & Gottleben (1957)	n = 44 DS (ages not specified) n = 472 AD* M _{CA} = 28.9 yrs for all participants (DS and AD)	Perceptual ratings: Assessments of speech articulation, voice, and stuttering	45% of DS were judged to stutter.
Also in Table A2.	*The AD group comprised AD individuals with other etiologies of mental retardation; all were residents of a training school.		
Rahovsky (1965)	n = 9 DS (2 M, 7 F) Range = 10;07–19;03 M _{age} = 15;10 (institutionalized) n = 18 DS (9 M, 9 F) Range = 9;04–19;07 M _{age} = 14;06 (noninstitutionalized)	Perceptual ratings: Fluency judgments (severity, incidence, and reactions of stuttering) by 10 graduate students enrolled in the study of speech and hearing	Stuttering identified in 35% of the institutionalized group and 19% of the noninstitutionalized group (both high and low verbal groups). A greater incidence of stuttering was found in females than in males.
Preus (1972)	n = 47 DS (21 M, 26 F; age 7+ yrs) NOTE: Participants were part of an Oslo-based day home for individuals with mental deficiencies.	Perceptual ratings and transcription: Analysis of stuttering and cluttering behaviors by 10 judges familiar with the individual with DS, based on a spontaneous speech sample Articulation testing: Articulation test used to screen for articulatory disorders	Stuttering on 5% of words was observed in 34% of individuals with DS, secondary symptoms were observed in 29.8% of individuals with DS, and cluttering was observed in 31.9% of individuals with DS. 52% of individuals with DS were judged to be stutterers. 46.8% showed no signs of cluttering. 10.6% had a pronounced tendency to stutter, and 31.9% were clutterers.

- What about the speech component in the disfluency?

DS & Speech development

Studies of both children and adults point to a higher than normal frequency of (inconsistent) articulatory errors, with substantial involvement of consonants (Brown-Sweeney & Smith, 1997; Bunn, Simon, Welsh, Watson, & Elliott, 2002; Kumin, 1994; Roberts et al., 2005; Rosin, Swift, Bless, & Vetter, 1988; Schlanger & Gottsleben, 1957; Sommers, Patterson, & Wildgen, 1988; Timmins et al., 2009; vanBysterveldt, 2009; vanBysterveldt et al., 2010).

Both the emergence and mastery of consonant phonemes in children with DS appear to be protracted processes, with substantial interindividual variability. The emergence of phonemes in the speech of DS children does not seem to follow the order of published norms for TD children (Kumin, Council, & Goodman, 1994).

Speech production in DS

Speech difficulties are not highly correlated with language or cognition, which may indicate that problems in speech are rooted in other factors, such as anatomy and motor control.

Some delay in the appearance of canonical babbling. Any such delay is modest compared with delays in gross motor skills.

Although peripheral factors such as anatomic anomalies are not likely to explain all aspects of the speech disorder in individuals with DS, the deviations may impose some limitations on articulatory performance (Beck, 2010; Bunton & Leddy, 2011; Leddy, 1999).

Review conclusions

The few studies reporting on prosody indicate that prosodic disturbance is a common feature of DS.

Disfluency (either stuttering or cluttering) is highly likely to occur in DS, but it is by no means a universal characteristic of the syndrome.

The diagnosis of cluttering, emphasizes the need to consider disfluency in relation to speaking rate Van Borsel and Vandermeulen (2008). Results on speaking rate in individuals with DS are mixed.

Speech task analysis

Semantic and phonological representations observed in a verbal fluency task.

Used the Predictive Cluttering Inventory (Daly, 2006), which was administered by 26 SLPs.

Perceptual ratings: Fluency judgments (severity, incidence, and reactions of stuttering)

Transcription: Analysis of stuttering and cluttering behaviors based on a spontaneous speech sample

Articulation test used to screen for articulatory disorders

Analysis of 7 disfluency categories for samples of spontaneous speech

Analysis of the relationship between speech disfluency and manual lateralization EPG assessment:

Verbal and manual motor production tasks at two levels of complexity

Analysis of conversational roles, conversational skills, and articulatory fluency

Determination of intelligibility and segmental accuracy & narrow phonetic transcription of recorded speech samples

Questionnaires

Examination of the production (using a nonword repetition task) and perception of word stress (using XAB discrimination task)

- Test situations: diversity

On command problem

Different level of focus on speech
production

Word / Non-word tasks

Conversational partner

Effect of changes in modalities is not
researched

- Other types of intellectual disabilities

Fragile X syndrome (FXS), a common form of intellectual disability, caused by large expansions of a trinucleotide (CGG) repeat in the X-linked FMR1 (fragile X mental retardation 1) gene (Verkerk et al., 1991).

This gene, which normally contains a repeat of 6–45 CGG trinucleotides, produces a protein (FMRP) critical for normal brain development. If the size of the CGG repeat expands into the ‘full mutation’ range (> 200 repeats), this usually leads to switching off the gene, and a gross deficit of FMRP (Pieretti et al., 1991).

The FMRP deficit is responsible for the intellectual disability and behavioural abnormalities associated with FXS (Loesch, Huggins, & Hagerman, 2004), through abnormal development of brain synapses (Comery et al., 1997; Irwin et al., 2000; Weiler & Greenough, 1999).

Females are usually less affected because of the presence of a normal FMR1 gene on a second, randomly switched off, X chromosome.

ASHA leader **

A family history of autism, mental retardation, fragile X syndrome, or learning problems

- **Physical signs**

long or protruding ears

long or wide forehead

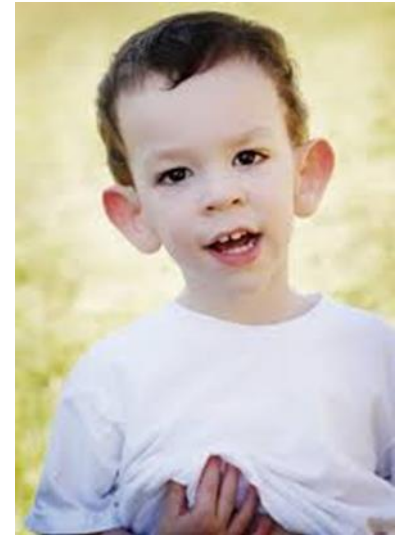
high, arched palate

- **Cognition**

males: moderate mental retardation, range from profound retardation to average intelligence

females: normal intelligence, with learning disabilities to mild-to-moderate mental retardation

males and females: visual spatial, attention, executive function, and math difficulties



** Roberts, J. , Hennon, E. A. & Anderson, K. (2003, October 21). Fragile X Syndrome and Speech & Language. The ASHA Leader

FXS (continued)

- Language

- difficulty maintaining a topic

- interested in social interactions

- word retrieval difficulties

- delays in vocabulary and syntax

- Adaptive behavior

- social anxiety and gaze aversion

- hyperarousal and hypersensitivity to stimuli

- stereotypic behavior such as hand flapping

- short attention span and hyperactivity

FXS (continued)

- Speech
 - perseverative and repetitive speech
 - reduced speech intelligibility (particularly in males)
 - rapid and uneven rate
 - phonological delays
- Oral motor
 - low muscle tone
 - motor planning and sequencing difficulties
 - tactile defensiveness

Figure 5a. Males with Full Mutation

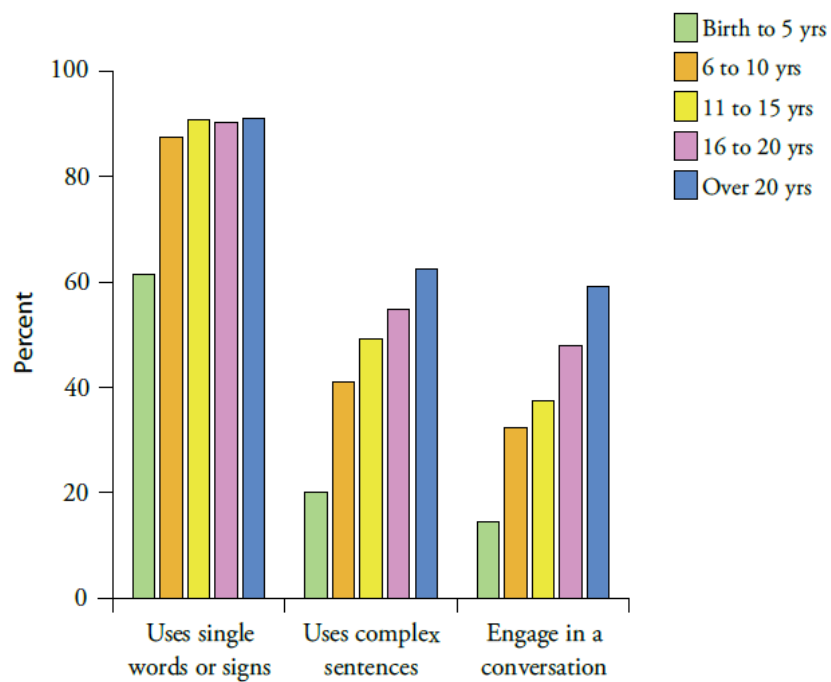
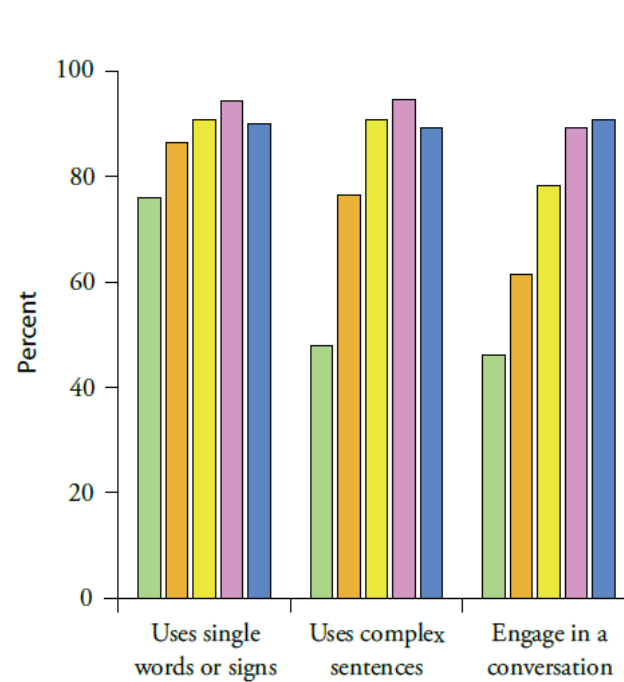


Figure 5b. Females with Full Mutation



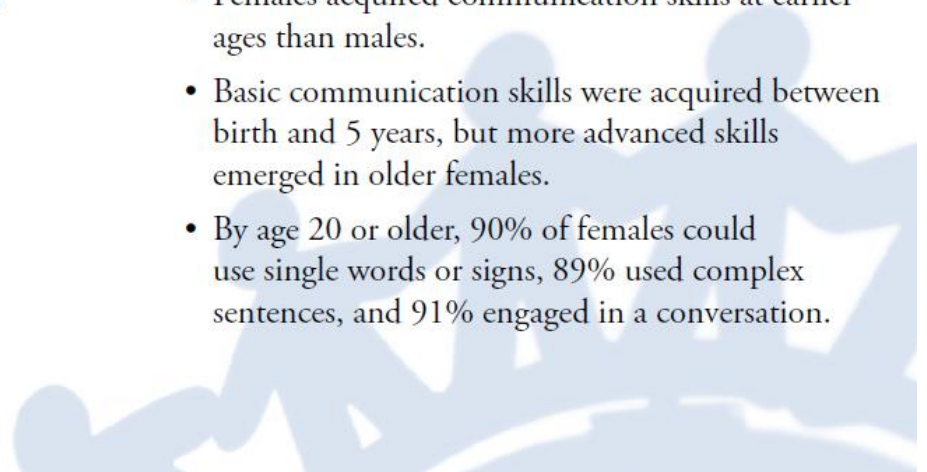
FXS communication

Highlights

- Males developed basic communication skills early, with more than 80% of males using single words or signs by 6 to 10 years of age.
- More advanced communication skills were slower to show change across the age categories.
- By age 20, the majority of males were able to use complex sentences (62%) and engage in a conversation (59%).

Highlights

- Females acquired communication skills at earlier ages than males.
- Basic communication skills were acquired between birth and 5 years, but more advanced skills emerged in older females.
- By age 20 or older, 90% of females could use single words or signs, 89% used complex sentences, and 91% engaged in a conversation.



http://www.fragilex.org/html/fx_survey.htm

FXS needs

primarily on studies of adolescents and adults with FXS.

Fluency and FXS

Van Borsel et al. listed five criteria which led them to conclude that the speech patterns of speakers with fragile X syndrome differed from those observed in developmental stuttering. The differences noted were:

- 1) distribution of type of dysfluency;
- 2) the class of word on which dysfluency occurred;
- 3) whether word length affected dysfluency;
- 4) number of times words and phrases were repeated; and
- 5) whether there were influences of material type on fluency (spontaneous speech, repeated material etc.).

The comparisons that van Borsel et al. (2007) made between participant groups were not for speakers of comparable ages. Comparisons with groups of corresponding ages support the opposite conclusion, namely the young speakers with fragile X syndrome show patterns similar to developmental stuttering, (Howell, 2008).

Intellectual disability and fluency

- Why do we know so little?
 - Exclusion criterium
 - Symptoms vs. Syndromes vs. Domains
 - Life is more important than communication

But what about:

- Communication is a human right (WHO, 2007)
- Participation & inclusion

Study Coppens, ... van Zaalen et al., 2013

N=28 adults (18–40 years; 16 men) with mild and moderate intellectual disabilities (IQs 40–70)

characterised as poorly intelligible by their caregivers

Dysfluencies were charted and classified.

Dysfluency profiles were related to mean articulatory rate and articulatory rate variability.

Coppens et al., 2013

- 75% of the MR participants showed dysfluency symptoms and 25% did not.
- 21% was cluttering, 29% cluttering–stuttering, 25% was cluttering with normal articulatory rate.
- No participant was classified as stuttering.

!!!!!!!!!!!!

Inclusion criterion:
bad intelligibility

Cluttering

- Speech, Motor, Planning, Execution, Rate, Language, Prosody
- Gropup or personal data???



Similarities & differences

Given that ability and disability are the products of multiple factors, this suggests that people with **similar static assessment** results, or with the **same diagnosis**, may actually perform very differently from each other in functional situations in the natural environment (Finke & Quinn, 2012).

- While two children may earn similar standard scores on **overall measures of IQ**, they are likely to show quite different profiles of performance when specific domains of development are examined (Fidler et al., 2008).

Down Syndrome

Table 2

Joint use of ICD-10 and ICF to document dimensions of developmental delay and disability in three children in

Dimension	Child	
	A	B
ICD-10: Health condition	Trisomy 21 Q90	Trisomy 21 Q90
ICF: Body function	b130 Energy and drive functions	167 Mental functions of language
ICF: Body structure	s4100 Heart anomaly	s240 Structure of external ear
ICF: Activities and participation	b1680 Language difficulties	d420 Walking difficulties
ICF: Environmental factors	e460 Societal attitudes	e585 Education and training services

*Simeonsson, Scarborough &
Hebbeler, 2006

6 DS children

Descriptives of the Six Participants

Child	Gender ^a	ICD-10 code ^b	Chronological age ^c	Mental age ^c	Expressive vocabulary age ^c	Receptive vocabulary age ^c
Jonas	M	Q90.0 T21 MN	4;9	1;9	1;10	1;5
Mary	F	Q90.0 T21 MN	4;4	1;11	1;6	1;5
Toby	M	Q90.0 T21 MN	3;1	2;0	1;9	1;6
Erna	F	Q90.0 T21 MN	3;9	2;6	1;8	1;7
Fenna	F	Q90.0 T21 MN	3;2	2;6	1;9	1;7
Nick	M	Q90.0 T21 MN	3;5	2;7	1;11	2;0

Note. ^aM = male; F = female. ^bT21 MN = Trisomy 21, meiotic nondisjunction. ^cAges are in years

Using ICF-CY to Uncover Strengths and Weaknesses in Communicative Competence of Children with Down Syndrome: Implications for AAC (Deckers, Van Zaalen, Van Balkom, Stoep & Verhoeven (under submission))

DS and Language

Domain	J	M	T	E	F	N
Language						
b16700 Reception of spoken language	-	-	-	*	+	*
b16710 Expression of spoken language-	-	-	-	-	*	
b16713 Expression of gestural language	*	-	-	+	+	*
b1672 Integrative language functions	-	*	*	*	*	-
d3101 Comprehending simple spoken messages	*	*	*	+	*	*
d330 Speaking	-	-	-	-	-	-
d331 Pre-talking	*	*	+	+	*	*
d3350 Producing body language	*	+	*	+	*	+

Using ICF-CY to Uncover Strengths and Weaknesses in Communicative Competence of Children with Down Syndrome: Implications for AAC (Deckers, Van Zaalen, Van Balkom, Stoep & Verhoeven (under submission))

Domain	J	M	T	E	F	N
Perception						
b1560 Auditory perception	-	*	-	*	*	*
b1561 Visual perception	-	-	-	+	+	+
b1564 Tactile perception	+	+	+	+	-	*
b1568 Perceptual functions (sensory integration)	-	*	*	*	-	*
b230 Hearing functions	*	-	*	*	*	*
b2302 Localisation of sound source	*	*	*	-	*	*
b235 Vestibular functions	*	*	*	*	-	*
b260 Proprioceptive function	+	*	*	*	*	*

Using ICF-CY to Uncover Strengths and Weaknesses in Communicative Competence of Children with Down Syndrome: Implications for AAC (Deckers, Van Zaalen, Van Balkom, Stoep & Verhoeven (under submission))

Children with DS seem to be just as different from one another as are all other children (Rogers, Gordon, Schanzenbacher, & Case-Smith, 2001; Deckers et al., in prep.)

And to my humble opinion this is as true for all other children with or without syndromes.

=> Serious implications for assessment

Fluency

- *“In most behavioural accounts it is helpful to explore the form of the behaviour one is interested inthen as a second step in understanding the behaviour, the stimuli that precede and follow it.....”*

(Starkweather, 1987)

Stuttering

Or as Ann would say:

In order to understand the moments of stuttering we need to know the cause (an impaired neural processing), the modulators (physiological arousal/cognitive demands) and the triggers (variable syllabic stress/linguistic complexity) (Packman, 2014)

Cluttering

Or as I would say:

In order to understand the moments of cluttering we need to know **the cause** (an uncontrollable fast output/rate), the **modulators** (physiological arousal/cognitive demands) and the **triggers** (phonological encoding processes/linguistic complexity).

Future research

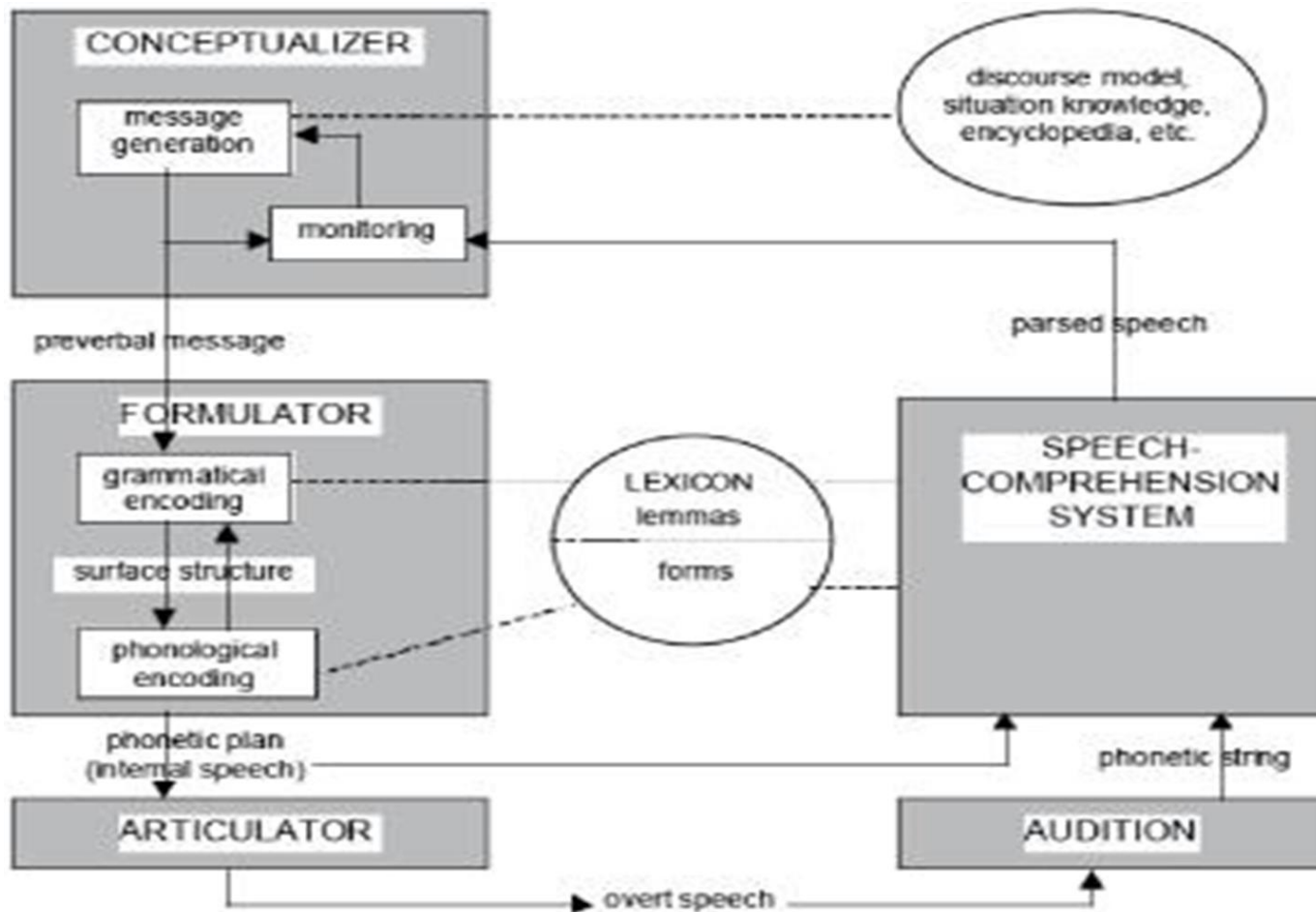
Research is needed that describes the communication skills of people with DS/FXS, examines the differences in communication skills from individuals with other forms of mental retardation, and assesses the sources of variability that may explain differences in communication functioning among individuals with DS/FXS.

Implications

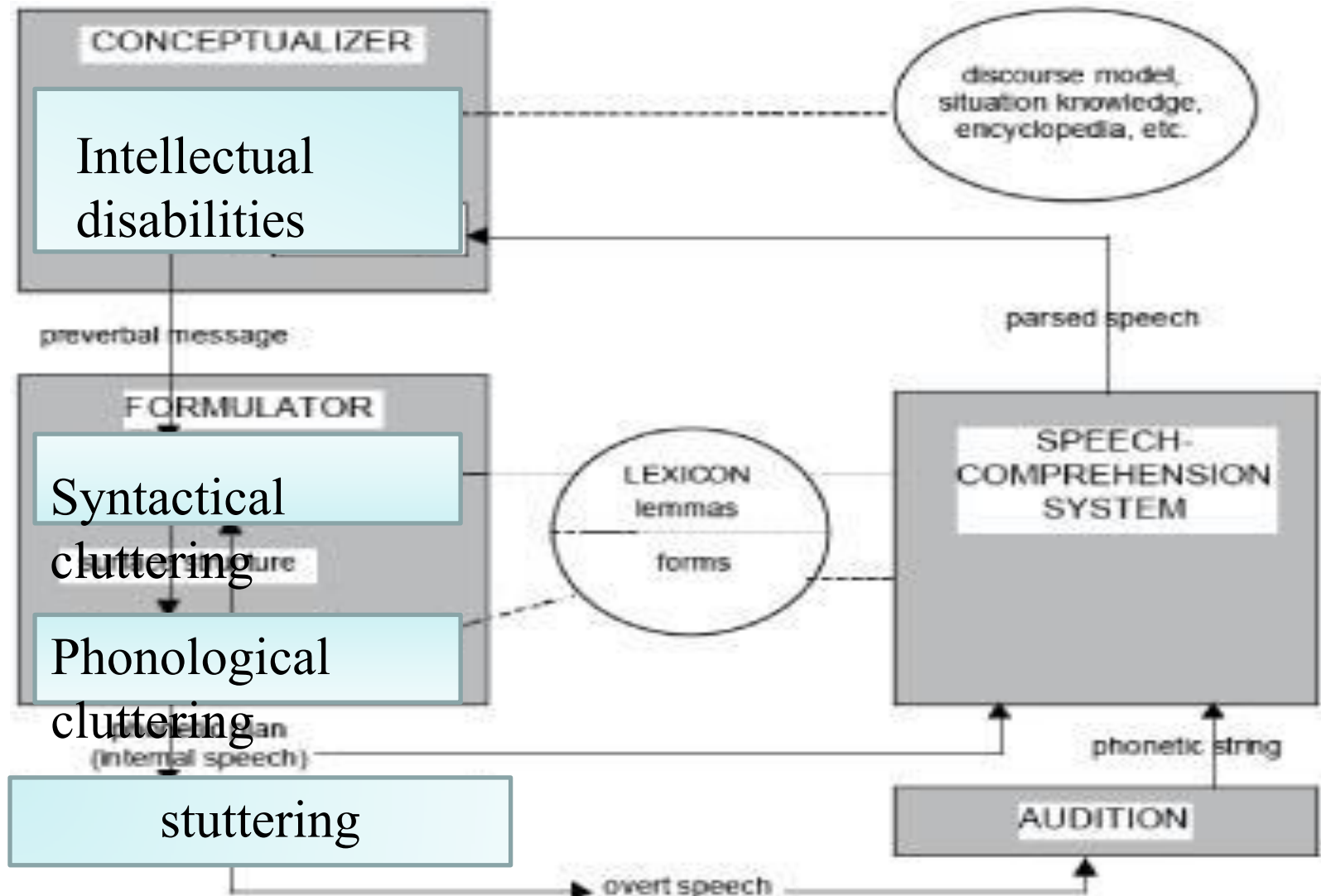
- Inclusion of participants with ID in research
- n = 1 study => model building => n=1 studysaturation

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Language production



Levelt's model



**THINK
BIGGER**



Fontys

**University of
Applied Sciences**