#### **STUTTERING**

## Speech control in children with Down syndrome

#### **Peter Howell**

Two areas where speech fluency problems arise, the motor system and the speech-language interface, are described and their implications for characterising the dysfluencies made by children with Down syndrome are discussed. Research topics are identified and implications for the treatment of speech problems in children with Down syndrome are drawn.

# Speech behaviours and their assessment in children with Down syndrome

Three types of problems with speech motor control are typically experienced by children with Down syndrome: 1) early articulation and phonological difficulties, 2) fluency disorders such as stuttering and cluttering, and 3) developmental aphasia-type problems. The first and second involve speech motor processes and the second and third involve interaction between the speech and language processes. This contribution details how motor factors affect speech and reports on the interface between language and speechmotor processes. At the end of each section, the fluency problems associated with these processes are presented and evidence whether children with Down syndrome are affected by these problems is discussed. Research questions that still need to be addressed are identified and the final section, some potential clinical implications are drawn.

## The speech-motor system in early articulation

#### Source

Sound production necessitates some air pressure movement. The principal source of that energy is the air in the lungs. This passes into the vocal cavities above the larynx. The escape of air from the lungs is controlled in different ways by structures in the larynx and, in some circumstances,

in the vocal tract itself. The energy to produce speech is referred to as the source of excitation. One major type of excitation is voicing (e.g. in vowels). In this mode, the vocal folds repeatedly open and close, which chops up the flow of air from the lungs<sup>[1]</sup>. A second type is voiceless energy, which can be produced when the vocal folds are held close together and a hissy stream of air escapes from the lungs into the vocal tract<sup>[1]</sup>, for example in the consonants 'p', 't' or 'k'. Although children with Down syndrome have breathing difficulties, which affects lung pressure, and motor coordination problems, which affects control of air escape from the lungs and manoeuvres of cartilage and tissues in the larynx, the impact these have on source production has received little attention. Therefore, related research questions are: how accurately can children with Down syndrome change from voiced to voiceless energy in connected speech, and is the sound that enters the vocal tract well-structured so that it leads to clear speech quality? Even superficial listening to the speech of children with Down syndrome suggests that the excitatory source is poor. Examination of details of the energy source is a major omission from the literature on children with Down syndrome.

Data are lacking on children with Down syndrome, but results on two adults with Down syndrome (one of each gender) have been reported which examined source characteristics<sup>[2]</sup>. High jitter rates (pitch perturbation) were reported, as were problems in articulating vowels.

#### Filter

The articulators above the larynx (commonly know as the vocal tract filter) change in position as speech is produced and this leads to the different sounds in languages. Thus, to produce the vowel /i/, a speaker lifts the tongue and pushes its tip forward in the vocal tract (close to the gum ridge behind the teeth, but not quite in contact). To produce the consonant /p/, the speaker brings the lips into contact (stopping the flow of air from the source) and then releases it[1]. Monophthongal vowels involve little or no movement of the articulators whereas consonants do. The sustained shape of the vocal tract can be estimated relatively easily for vowels, as vocal tract resonances (formants) relate to the two coordinates of movement (tongue height and its forward/backward position). There have been no reports about how children with Down syndrome compare to controls in vowel production, although there has been a study on adults<sup>[2]</sup>.

MRI scans show clearly that children with Down syndrome have different shaped vocal tracts from controls and specify where in the tract these differences occur<sup>[3]</sup>. The main difference is that the front part of the roof of the mouth is reduced in children with Down syndrome. The amount of data is limited at present and more exploration is needed, hopefully in the form of longitudinal scanning data for both genders. This should be used to examine how the anatomical differences affect vowel articulation. Currently MRI scans have only been used for anatomical measurements, but it should be possible to obtain scans while children with Down

syndrome and controls hold the vocal tract shape for vowels. Coordination between the articulators in consonant production can be measured using physiological or acoustic techniques. Acoustic measures are preferred as they are not invasive<sup>[2]</sup>. Howell, Anderson and Lucero describe techniques that are appropriate for use with children with Down syndrome<sup>[4]</sup>.

## The interface between language and the motor system

There are two main views about how the motor system receives language input from the language system that can lead to fluency problems. One considers that fluency problems are a result of impaired (error-prone) language processing and that the motor system responds passively to whatever language input it receives. It has been proposed that speakers become aware of these errors by listening to their own speech (monitoring). If errors occur, the speaker interrupts and repairs them, which leads to aspects like whole or partword repetitions and pauses that are seen when there are fluency problems<sup>[5,6]</sup>.

According to this view, if children with Down syndrome have acute language problems, they should make more linguistic errors of all forms than typicallydeveloping children. Such groups have not been compared systematically for: a) relative incidence of language errors, b) whether there are specific components of language processing that are particularly error prone in children with Down syndrome. Children with Down syndrome have hearing problems which would affect how well they can monitor their overt speech output. There has been no study which has assessed monitoring of speech output in children with Down syndrome compared to normal-hearing control children or controls with similar hearing impairments to children with Down syn-

The second alternative argues that the dysfluency phenomena mentioned earlier arise because the linguistic system has to provide the output at a particular time for the speech sequence to be produced fluently<sup>[7,8]</sup>. If the language representation does not produce the output to the speech motor system at the required time, speakers hesitate or repeat words until the

material is ready and they can proceed, accounting for the same dysfluencies as Levelt, in a different way. This account does not assume there is monitoring.

#### Stuttering

Stuttering has been characterised as involving fluent speech with interspersed dysfluencies. It can also have associated secondary characteristics that occur while speech is taking place (e.g. foot tapping or facial grimaces) and affect wider aspects of behaviour. The overlap in the type of dysfluencies seen in typically developing and stuttering children makes diagnosis difficult (see REF 9 for a review). However, it appears that part-word repetitions, prolongations and word blocks are features that are commonly seen when stuttering persists. Although there is a wealth of research evidence that indicates motor and linguistic factors affect stuttering in typically-developing children, the data on stuttering in children with Down syndrome are sparse and usually report prevalence rates of stuttering in children with Down syndrome and some performance differences between children with Down syndrome who stutter and those who do not<sup>[10]</sup>. Studies are needed which look separately at linguistic and motor influences on stuttering. Preus reported results on 47 speakers with Down syndrome out of 67 whose speech was deemed intelligible to a panel of judges<sup>[10]</sup>. When whole-word and part-word repetitions and prolongations were examined, 46.8% were judged stuttering. If whole-word dysfluencies were excluded, the frequency of stuttering reduced to 34%. Preus considered that secondary symptoms such as body movements, avoidance and postponement are signs of awareness of stuttering and indicative of a more advanced form<sup>[10]</sup>. When only these signs were included, 29.8% of the sample was still considered to stutter, which suggests that stuttering in its advanced form can occur in children with Down syndrome. Some questions that remain are how stuttering originates, how it compares with typically-developing controls, how it develops, whether it progresses at the same rate as in typicallydeveloping controls. Longitudinal work on children with Down syndrome would be best to answer these questions.

In a later examination of the earlier data, Preus reported that 34% of the

group of children with Down syndrome had symptoms of stuttering and 31% had symptoms of cluttering [11]. Cluttering is a pattern of speech closely allied to stuttering. For instance, cluttering appears as the same categorical disorder as stuttering in DSM-IV[12]. Other authors (most recently Ward<sup>[13]</sup>) argue that stuttering and cluttering are distinct. Some of the defining characteristics that set cluttering apart from stuttering are: 1) poorly organised thinking; 2) short attention span and poor concentration and 3) lack of complete awareness of the problem and 4) high speech rate. Characteristics like 1) and 3) are difficult to measure. At a recent conference, Howell and colleagues showed that nine samples of clutterers from a longitudinal database of clinical cases referred for fluency disorder could not be distinguished from speakers who stuttered but who later went on to recover. It would therefore seem preferable to assess cluttering along with stuttering in children with Down syndrome and not as being distinct from it.

## Developmental aphasia type problems

Telegraphic speech refers to loss of function words. Speakers with Down syndrome have been reported to have telegraphic speech<sup>[14,15]</sup>. Loss of function words is often associated with dropping of morphologically-determined inflectional endings. Eadie, Fey, Douglas and Parsons showed this in a study they performed that compared children with Down syndrome, controls (matched on mean length of utterance) and children with specific language impairment<sup>[16]</sup>. Problems were reported for both the children with Down syndrome and those with specific language impairment for word-final inflections. Related findings were reported by Vicari, Caselli and Tonucci for 15 children with Down syndrome and 15 mental-agematched controls[17]. They reported no difference in lexical development, but significantly poorer ability by the children with Down syndrome in terms of morphological processing.

#### Clinical implications

#### Early articulation

It needs to be established whether preventing problems at the early stages of

speech development prevents subsequent speech and language problems, i.e. does remediation dealing with early articulation problems reduce the chances of the development of stuttering? In terms of the two components of speech that need to be controlled (source and filter), speechlanguage therapists have vast experience in teaching breathing control (source), although comparatively little has been done with very young children, and no reports could be found on children with Down syndrome. Tongue shortening has been tried as a way of dealing with vocal tract configuration issues in people with Down syndrome (and also as a treatment for stuttering)[18]. This seems a radical solution and there are alternatives. For instance, speakers learn to adapt to new articulatory configurations (e.g. after trips to dentists) and procedures for teaching compensatory strategies like these are a less dramatic alternative that should be examined first. More data are required

on vowel and consonant production and speech errors at these early stages before clinical implications can be drawn.

#### Fluency disorders

Cooper suggested a behavioural modification programme for treating people with Down syndrome who stutter<sup>[19]</sup>. Instead of modifying or shaping feelings that may inhibit the development of improved fluency, the focus should be on concepts and explanations that are appropriate to the child's conceptual and linguistic skills. However, for these methods to work the child needs to be aware that he/she stutters, which may not always be the case with children with Down syndrome. There are methods that also work whether or not the child is aware of stuttering: for example, operant conditioning procedures, as verbal conditioning can occur without awareness<sup>[20]</sup>. One treatment that is widely used with young children is the Lidcombe programme. If the speech of children with Down syndrome who stutter develops in the same way as in otherwise typically-developing children who stutter, later forms of treatment would need to be developed for treating stutter in their speech (see REF 21 for a potentially suitable procedure that has been used in experiments).

### Developmental aphasia type problems

In terms of the symptoms that show some similarity with aphasia, it is primarily necessary to establish whether this supposed similarity actually obtains. If it does, then the techniques which have been successful with children with this disorder who do not have Down syndrome could be employed with children with Down syndrome. However, again more data are required about the supposed similarity before clinical implications can be drawn.

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Peter Howell is at University College London.

**Correspondence to** Peter Howell, Department of Psychology, University College London, Gower Street, London WC1E 6BT, UK • e-mail: p.howell@ucl.ac.uk

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