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Speech production in children with Down's Syndrome: the effects of reading, naming and imitation.

Abstract

People with DS are known to have difficulties with expressive language, and often have difficulties with intelligibility.   They often have stronger visual than verbal short-term memory skills and therefore, reading has often been suggested as an intervention for speech and language in this population.   However, there is as yet no firm evidence that reading can improve speech outcomes.   This study aimed to compare reading, picture naming and repetition for the same 10 words, to identify if the speech of 8 children with DS (aged 11-14 years) was more accurate, consistent and intelligible when reading.   Results show that children were slightly, yet significantly, more accurate and intelligible when they read words compared to when they produced those words in naming or imitation conditions although the reduction in inconsistency was non-significant.   The results of this small scale study provide tentative support for previous claims about the benefits of reading for children with DS.   The mechanisms behind a facilitatory effect of reading are considered, and directions are identified for future research.

Introduction

This study investigates the effect of reading aloud on the speech production skills of children with Down's syndrome (DS).

DS is the most common genetic cause of learning disability (Patterson and Lott, 2008) presenting in 1.08 of every 1000 live births in England and Wales (Morris and Alberman, 2009; p1).   DS is usually caused by the presence of an extra chromosome at location 21 (Trisomy 21) (Sherman *et al.*, 2007) either in all cells, or, for individuals with mosaic Downs Syndrome (around 3% of all those with DS), in only some cells.   DS is expressed in a physical as well as a cognitive phenotype (Patterson, 2007).   Intellectual impairment is characterised by a slow cognitive processing rate, deficits in verbal working memory and delays in language development (Silverman, 2007). The mean intelligence quotient (IQ) of the DS population is 50, with a delayed rate in the development of cognitive skills (Chapman and Hesketh, 2000), so that people with DS are typically considered to have mild to moderate learning disability (Roizen, 2007).

Speech and expressive language are areas of particular concern in the development of children with DS, and Roberts *et al.* (2007) suggest that speech production and expressive vocabulary are further delayed in DS than in typically developing children of a similar mental age.   Within the DS population, both speech and expressive language are at a lower level than comprehension and nonverbal intelligence (Laws and Bishop, 2003).   The majority of children with DS experience difficulties with speech intelligibility (Cleland *et al.*, 2010), with up to ninety-five per cent of parents reporting that their children with DS either 'frequently' or 'sometimes' have difficulty being understood by others (Kumin, 1994).   Barnes *et al.* (2009) confirm that children with DS have significantly lower intelligibility scores than typically developing children of similar non-verbal developmental age.

Part of the explanation for difficulties with intelligibility lies in the phenotypic characteristics of children with DS, which include physical impairments that are likely to impact on their speech. For example, approximately two thirds of children with DS are affected by sensory-neural or conductive hearing loss (Roizen, 2007).   Additionally children with DS have atypical oral structures and functions (Barnes *et al.*, 2006; Roizen, 2007), such as a small oral cavity and narrow, vaulted palate, as well as hypotonia, and differences in nervous innervations (Venail *et al.*, 2004).   However, recent literature has concluded that these physical characteristics do not entirely account for the speech deficits experienced in DS (Dodd and Crosbie, 2005; Cleland *et al.*, 2010).

One reason for suspecting underlying difficulties, beyond oral motor differences, is that the phonetic accuracy of children with DS increases when words are imitated compared to when they are spontaneously produced (see e.g. Dodd 1976; Lennenberg, 1967).   According to the speech processing model of Stackhouse and Wells (1997, p45), better repetition than naming skills indicates that lower level articulatory processes of motor execution are not the primary source of impairment, and that speech difficulties are more likely to arise due to difficulties with stored phonological representations or motor programmes.

An additional reason for assuming a more central deficit in speech production comes from the inconsistent productions typically observed in children with DS.   Dodd and Thompson (2001) indicate that children with DS are more than 60% inconsistent when naming the same pictures

three times within an assessment session.   Dodd and colleagues (e.g. Crosbie *et al.*, 2005)

have also demonstrated that using a 'core vocabulary' approach, which targets consistency,

rather than correct production per se, can increase the number of consonants produced

accurately.   Again, inconsistency suggests the involvement of central, as well as peripheral,

speech mechanisms. Furthermore, Kumin (e.g. 2006), from a large scale survey of parents,

demonstrated that the majority of children with DS showed signs of dyspraxia, such as

inconsistent productions.

Difficulties with verbal short term memory have been proposed as one central mechanism that

might account for the speech difficulties observed in DS. Difficulties with verbal short term

memory have been linked to problems forming accurate phonological representations, and

studies have consistently shown that children with DS experience a selective impairment to

verbal short term memory (e.g. Laws, 2002; Baddeley and Jarrold, 2007; Frenkel and Bourdin,

2009). As information must be processed in short term memory before its long term

representation can be assembled and stored (Gathercole and Baddeley, 1993), a deficit in short

term memory could lead to incomplete or inaccurate long term phonological representations of

words.   This difficulty with compiling accurate phonological representations could in turn lead

to both the inconsistency observed in speech production, and a pattern of better repetition

than naming skills.   Indeed, recent evidence has shown that DS children's verbal short term

memory skills are related to their speech production (Laws, 2004).

Whilst verbal short term memory has been shown to be impaired in children with DS, visual

working memory often functions at a higher level (e.g. Laws, 2002; Baddeley and Jarrold, 2007; Frenkel and Bourdin, 2009).   In order to benefit from these strong visual memory skills, whilst minimising the role of verbal short term memory, reading has been promoted as an intervention for speech difficulties in children with DS.   The suggestion for intervention via reading takes as its starting point Buckley and Bird's (1993) observations of children with DS following a reading program.   Data on spoken and written word acquisition over the program is provided for a single case, and the paper concludes that 'reading practice improves phonology and articulation' (p.36).   More recently, Dodd and Crosbie (2005) suggest that learning the visual form of a written word avoids verbal short term memory deficits, as the visual form can be paired to its phonological form for spoken output.   They note that this process would 'allow a lexical representation to be formed and provide a map for consistent phonological output' (p.240).

However, not all evidence points towards a causal relationship between reading and improved speech outcomes.   Laws (2010) reports a two year study comparing a group of emerging and non-readers with a group of competent readers, all with DS.   In measures taken over two years there was a non-significant trend toward higher scores for percentage of sounds produced correctly in the 'competent reader' group.   However, no interaction between improvements in speech scores and reading group (non-readers and competent readers) was found. The study concluded that there was no evidence for a link between reading and speech level.   Additionally, Laws (2010) reviewed the literature regarding reading as an intervention for speech development.   Whilst several studies in the review 'showed associations between

reading and other functions, there was little clear evidence that reading was the driver in these relationships' (p.153).

Thus, whilst reading is believed to be a useful route towards improved speech production for children with DS, firm empirical evidence for this claim is somewhat lacking.   The present study aims to further investigate the impact of reading on the speech of children with DS by comparing their productions of the same words when they are read, named from pictures, or imitated.   The research targets three areas of speech production known to be impaired in children with DS, namely their accuracy of phoneme production, their consistency when the same words are repeated, and how intelligible their speech is to listeners.   Thus, the research questions to be addressed are whether the speech of children with DS is more accurate, more consistent and more intelligible when words are read, compared to when they are imitated or named from pictures.

Method

<u>Participants</u>

All participants were recruited through email and letter advertisements distributed through a national charity.   Initially interested parents completed a form to confirm that their children were between ten and fourteen years, had no major medical conditions or additional disabilities and were currently uninvolved in other research.   Children were also required to be able to read the sentence: "The boy dropped the knife". A questionnaire was completed by a parent of each participant detailing further information including type of education, perceived intelligibility sensory impairments and language background.   Eight children (two male and six female) with Down's syndrome took part.   Their

ages ranged from 11 to 14 years.   One participant had a diagnosis of mosaic DS, and all others

full trisomy 21.   Full details for each participant are shown in appendix A.   Two further

participants were recruited, but data was excluded for one boy due to incidental loss of data,

and for another due to a lack of ability to manage reading tasks.

All participants were required to speak Standard Southern British English (SSBE) as their first

language although three children were reported to have second languages (French, Urdu and

Portuguese).   All participants had a mild visual impairment (astigmatism or long-sightedness).

Only three participants were reported to have normal hearing while four had mild hearing loss

and one wore permanent hearing aids as a result of mild to moderate hearing loss.

As per the ethical approval received from City University London Research Ethics Committee, all parents

received an information letter detailing the study and signed consent for their child's participation at

least two weeks prior to the testing date. At the beginning of the testing session, participants read a

consent letter together with the experimenter and coloured in a smiley face if they wished to

participate) . All children seen for testing consented to participation in the study.

Stimuli

Ten single words were chosen which were appropriate for reading, naming and imitation tasks,

and which are typically early acquired in speech and reading.   Thus the ten words (shown in

appendix B) were imageable nouns, representing highly-recognisable items (e.g. glove, jam, and

bath), had a regular orthography, and were either one or two syllables in length.   All

consonants in the English repertoire were used once in the word list (giving a total of 24

consonants).   Two words with initial two-consonant clusters and two with final two-consonant clusters were included, as cluster reduction is known to be a common error pattern for children with DS (Cleland *et al.* 2010, 90).   For the reading condition, words were printed onto flash cards (font: Calibri, size: 48).   For the picture naming condition a set of coloured picture cards representing the words were drawn.   For the imitation condition, words were recorded by a female with an SSBE accent.

Procedure

Testing was completed within one twenty to forty minute session depending on each participant's attention span and need for breaks.   All participants were tested on an individual basis in their home environment.   A quiet and non-distracting room was requested by the experimenter although in some sessions low levels of background noise could not be eliminated.   Parents were occasionally present but were requested not to interrupt the session.   The experimenter ensured that necessary sensory aids (spectacles and hearing aids) were functioning before initiating the testing session.

The participants were tested while sitting at a table with the experimenter sitting opposite or diagonally across from them, so that the distance between child and experimenter was approximately 30 to 50 cm.   Sessions were recorded with a Marantz PMD660 recorder, which was placed on the table approximately 30 cm from the child's mouth, although minor variations occurred due to accessible space and movement of the child.   Speakers were placed at approximately 40 cm distance from the child for presentation of the words in the imitation

condition.   Speaker volume was tested and adjusted to the child's hearing level by asking them to repeat a practice word (different from those in the main trial) at the beginning of the session.   The established volume was then maintained throughout testing.

Words were presented in the same order for each participant in every condition.   However, conditions were counterbalanced between participants using the simple counterbalancing method.   Children produced all words in one condition, before moving onto the second and then the third condition.   They were then offered a break, and given the opportunity to play with a puppet or ball.   This procedure was repeated three times, so that three repetitions of each word in each condition were obtained.

Children were told that they would play some 'word games' with the experimenter.   At the start of each condition, two practice words (separate from, but of a similar complexity to, those in the main study) were administered to ensure that presentation was adequate and that children understood what was required of them before moving on to the test stimuli.   The distance that word and picture cards were held from the child was also established through practice items and then maintained in testing. This distance ranged from 15 to 25 cm.

In the reading condition, participants were shown the flash cards of written words and asked to read them aloud.   No further cues were given.

In the naming condition children were shown the pictures representing the stimuli items, and

asked to tell the experimenter what they saw.   No further cues were given unless the child

made a naming error due to image ambiguity.   Ambiguous naming occurred with three

pictures, where yellow was mistaken for 'paint', stick for 'twig', and ship for 'boat'. In these

cases, one question was initially used to elicit the correct word (e.g. if a child said 'paint' for

yellow, the examiner asked 'what colour is it?') although this question could be repeated and

varied up to three times.   These questions were developed in advance and used consistently.

In the imitation condition, recorded tracks of the stimuli items were played from Windows

Media Player with amplification from two Labtec speakers. Children were told to say the words

back to the experimenter and no cueing was used.   Whilst this task is essentially a real-word

repetition task, the term 'imitation' is used in this paper, to avoid confusion with the term

'repetition', which will be used to mean the number of times an item was presented.

Analysis

In order to assess accuracy and consistency of productions, phonetic transcriptions were made

of children's responses in all three conditions.   The audio recordings of all testing sessions

were segmented into tracks, each containing a single production of a word.   All tracks were

then pseudo-randomised and burnt to a CD.   Ten per cent of productions were included twice

so intra-transcriber reliability could be calculated.   All items were narrowly transcribed by a

trained phonetician with experience of a wide range of clinical speech, who completed

transcriptions in an unmonitored environment with no limit on time or number of listenings.

The transcriber was blinded to the participant number, condition, repetition number and

attempted word.   Intra-transcriber agreement was 90.04%.   A second transcriber

independently transcribed 120 randomly selected productions.   Inter-rater reliability was

87.36 %, calculated on a segment-by-segment basis.

Accuracy

The accuracy of productions was assessed by calculating the percentage of consonants

produced correctly (percentage consonants correct, PCC).   PCC was calculated by comparing

the phonetic transcription of each repetition to the phonetic transcription of the of the

Standard Southern British English production.   Any consonants missing or in error were scored

as incorrect, with allowance made for accent features of southern English, such as glottal

replacement, and labiodental productions of /r/ or 'dental' fricatives.   There is debate about

whether common errors, possibly due to anatomical differences, such as dental or lateral /s/

are best scored as correct or incorrect; here such errors were scores as incorrect.   PCC for

each condition was then calculated using the formula: PCC = (consonants in error ÷ consonants

attempted)*100.

Inconsistency

Consistency comparisons were carried out only on words repeated three times within a

condition, following the procedure in Dodd and Thompson (2001). Thus, transcriptions were

judged for consistency on a phonemic basis and a score of zero was attributed where all three

word repetitions within a condition were consistent.   In cases where any of the three word

repetitions was produced differently, a score of one was given.   A percentage of inconsistent

production was then calculated as follows: inconsistency = (number of words produced

inconsistently ÷ total number of items produced three times) * 100.   A higher percentage

therefore represents less consistency between word productions.

Intelligibility

Intelligibility was assessed by a group of twelve female Speech and Language Therapy

Undergraduate students in their fourth and final year of study.   Raters ranged in age from 21

to 44 years (Mean = 27.75, SD = 8.4).   All raters had completed and passed modules in

phonetics and child speech development, and seven stated that they had experience with

children with DS.   Ten raters reported their first language as English with two reporting Urdu

and Ukrainian as their first language.   All listeners had lived and studied in the London area for

at least three years.   None of the participants had a speech difficulty while eleven reported

normal hearing.   One participant described mild right ear hearing loss.   However, her ratings

did not differ significantly from other participants so her were included.

The first production of every word from each condition was rated for intelligibility.   This was in

order to assess the effects of each condition directly, rather than any practice effects that might

be present in the second or third repetitions, and to make the rating task manageable for

raters. Ten per cent of productions were rated twice so intra-rater reliability could be

calculated.   Raters were required to complete five practice items before beginning to rate

test items, and then a total of 264 tracks were rated in a random order, with raters blind to the

participant and condition. Raters were provided with the attempted word in orthographic form

and were asked to compare the child's production to their knowledge of a typical SSBE

production of this word.   They then rated the child's production between 1 and 5, where 1

equalled 'completely unintelligible' and 5 equalled 'completely intelligible'.   After each track

was played, raters had 4 seconds to respond by circling the appropriate rating on paper.   Two

breaks were given at regular intervals during the session.   Raters were allowed to request one

repeated playing of a track.   Following others in the literature, mean scores were calculated

per listener and condition, thus treating the data as an interval scale (see e.g. Jamieson 2004;

Norman, 2010; Carifio and Perla, 2007, for more on this issue).

In order to calculate reliability, scores were counted as consistent when they varied by only one

point on the rating scale. Intra-rater reliability scores ranged from 64.58 % to 95.84 % with a

mean of 86 %.   Inter-rater reliability was 88 %.

Results

<u>Accuracy</u>

The accuracy of productions in the three conditions was investigated by entering the

Percentage Consonant Correct scores into a linear mixed effects model (Baayern, 2008).   Such

models are considered suitable for data sets with unbalanced and missing data.   The initial

model was fitted with the predictors <u>condition</u> (reading, naming, imitation) and <u>repetition</u> (1st,

2nd, 3rd) plus their interaction, and random effects for <u>participant</u> and <u>word</u>.   However, there

was no significant interaction between the predictors, so results are given for the model

without this interaction.

As shown in figure 1, there was a significant effect of condition ($\chi 2$=7.39, df=2, p<.05). Planned comparisons indicate that accuracy when reading (68.9% PCC) was significantly better than when naming (60.1% PCC) (p<.05), or imitating (61.2% PCC) (p<.05), but that there was no significant difference between naming and imitation (p= 0.64).

There was also a significant effect of repetition ($\chi 2$=7.12, df=2, p<0.05), again shown in figure 1. Accuracy in the second (65.2% PCC) and third (66.2% PCC) repetitions was higher than in the first repetition (58.4%PCC, p<.05), but there was no significant difference between accuracy in the second and third repetitions (p>.05). Individual results for all measures are shown in Appendix C.
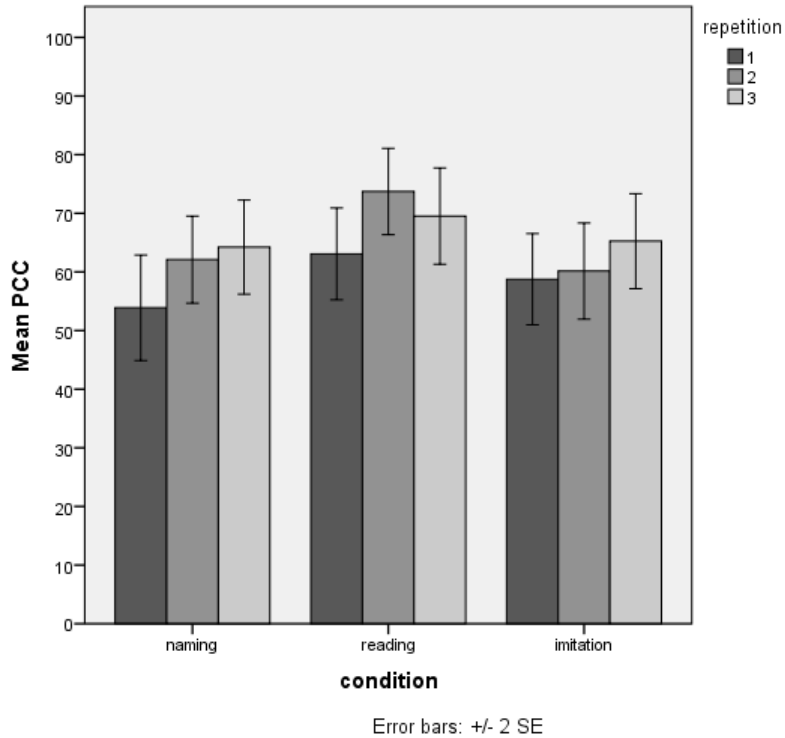
Figure 1 – Mean Percentage Consonants correct by condition and repetition.

Whilst a full analysis of error patterns is beyond the scope of the current paper, children

presented with both developmental (e.g. cluster reduction, gliding) and atypical (e.g. initial

consonant reduction) patterns, as well as some not easily classified.

Inconsistency

Inconsistency results are based on data from 7 participants, as one child produced three

repetitions on too few occasions.   Results are shown in figure 2.   Mean scores for

inconsistency were compared using a repeated measures ANOVA with the factor condition

(reading, naming, imitation).   Despite a numerical reduction in inconsistency for the reading

(45.8%) over the naming (62.7%) and imitation (59.1%) conditions, there was in fact no

statistically significant difference between conditions ($F_{(2,12)}=1.256$, $p>.05$), and a great deal of

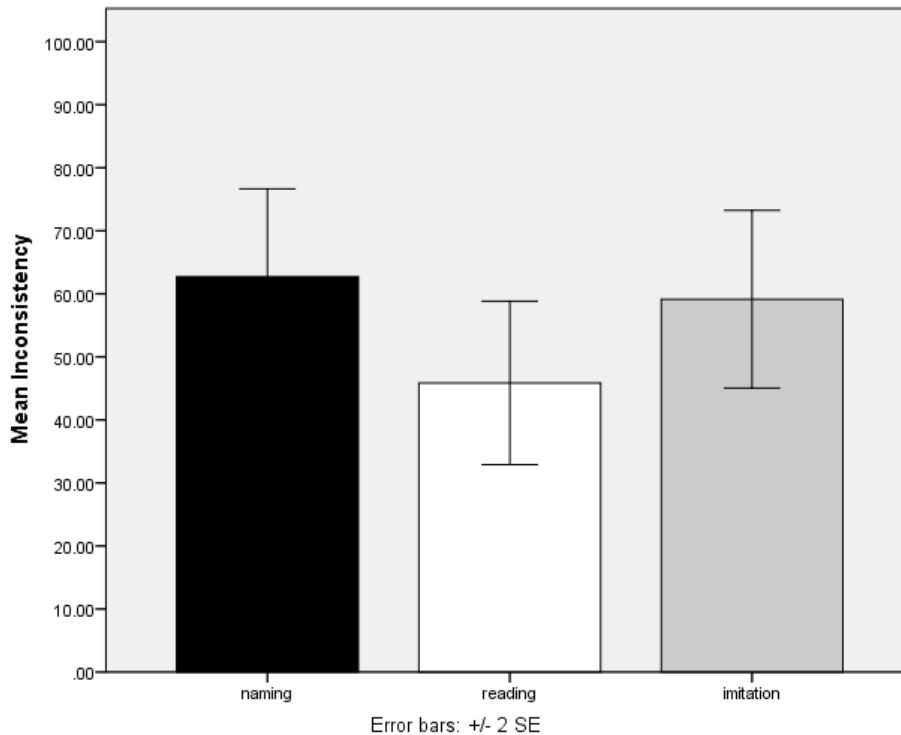individual variation was present.

Figure 2 Mean inconsistency by condition

<u>Intelligibility</u>

Results for intelligibility are shown in figure 3.   The overall effect of condition on intelligibility

ratings was tested firstly using a Friedman's test, which was statistically significant ($X^2(2)=$

22.17, $p<.01$).   Planned comparisons were completed using Wilcoxon Signed rank tests, which

show that productions from the reading condition (mean=3.74) were slightly but significantly

more intelligible than those from the naming condition (mean=3.66) ($z=-2.86$, $p<.01$) or those

from the imitation condition (mean = 3.27), ($z=-3.06$, $p<.01$). Furthermore, naming productions

were also rated as more intelligible than imitated productions ($z=-3.06$, $p<.01$).
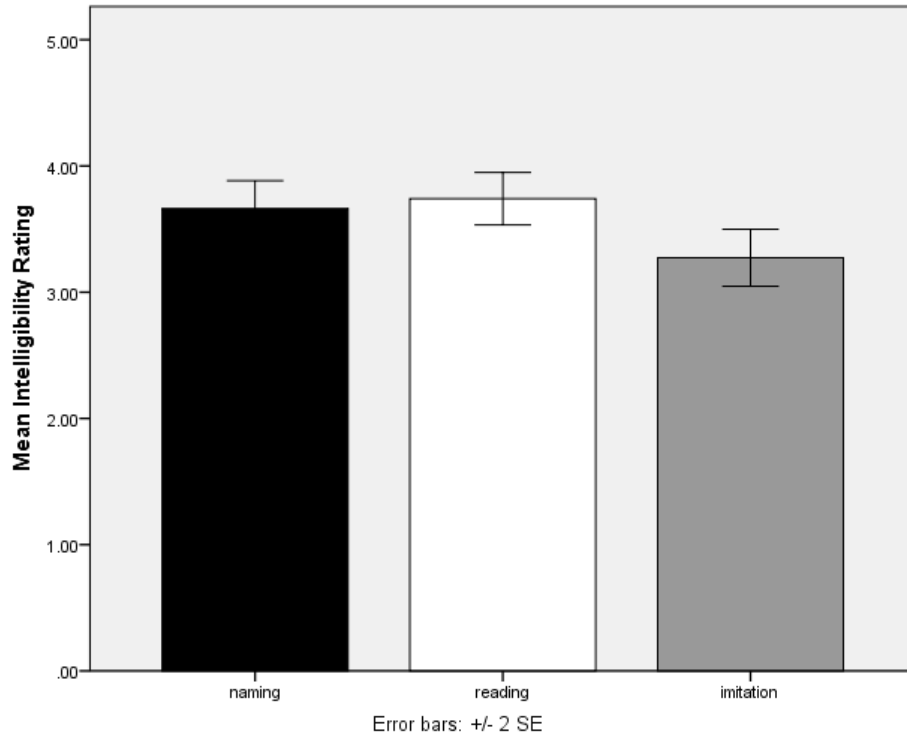
Figure 3. Mean intelligibility ratings by condition

Discussion

This research investigated whether reading aloud can improve the speech production of

children with Downs Syndrome.   Children produced three repetitions of the same ten words in

three conditions (naming, reading and imitation).   Productions were scored for accuracy

(percentage consonants correct), and inconsistency across the three repetitions, and were also

rated for intelligibility.   This study investigated a relatively small number of children and

words, and thus replication with a larger sample size would be a fruitful next step in order to

further investigate the role of reading for children with DS.

Accuracy

In answer to our research question, children with DS were more accurate in their productions when they read words, compared to when they named or imitated those words.   One partial explanation for this result is that the printed word allows children to rely on their strong visual skills, bypassing inaccurate or incomplete stored motor programmes, and difficulties with short-term memory.   In terms of a psycholinguistic framework (such as Stackhouse and Wells, 1997), reading allows access to a motor programme from the orthographic form, via either the phonological representation or semantic representation.   Naming, on the other hand, requires children to rely on accessing a stored motor programme via their semantic representation. Imitation allows children to access a motor programme directly from the phonological representation of the auditory stimuli, or to access a stored motor programme.   It seems likely then that naming performance was impaired (relative to reading) due to accessing inaccurate motor programmes, whilst imitation was impaired either for the same reason, or because of a difficulty in storing words in short term memory.   These results might support a more phonological, rather than oral motor or motor programming, account of the speech difficulties in DS.   A more detailed discussion of the facilitatory effect of reading, and the potential routes used to read, will be provided at the end of the discussion.

It is also noteworthy that accuracy improved after the first repetition, but not between the second and third repetitions.   This was the case in all conditions, and there was no interaction between repetition and condition.   This finding suggests that producing words once, in any

condition, had a facilitatory effect on later productions.   This finding could perhaps be interpreted as children having to form new motor programmes for relatively unfamiliar words during the first repetition, which were then used in later repetitions.   Although common words were used, it is possible that they were not used productively by the children in the study before the start of testing, so no stored motor programmes were available.   Further research could ensure that target items are in the expressive vocabulary of participants, via parental report.

Although conditions were counterbalanced, children completed one production in all conditions before moving onto the next set of productions.   Thus, they had produced each word three times before producing it again in the same condition.   This design was important to the present study, but makes it difficult to tease apart the contribution of repetitions across conditions.   Additional testing would be beneficial, using a design where, for example, all reading productions were completed before moving on to all naming productions, and so on.

In general, accuracy of all productions, as measured by Percentage Consonants Correct, was high.   Accuracy was between 60% and 69% depending on condition (although there was a good deal of individual variation as shown by the standard deviations). Scores in the 50-64% range indicate moderate to severe speech sound disorder, whilst those from 65-85% indicate a mild-moderate disorder according to Shriberg and Kwiatkowski's (1982) original severity classification.   Although such classifications are based on conversation samples, Garret and Moran (1992) have shown high correlations between PCC calculated from single words and

conversational samples.

In general the results for accuracy of naming (60.1% PCC) are in line with others in the literature using single word naming tests with children who have DS.   Cleland *et al.* (2010) report PCC scores from the phonology subtest of the DEAP as between 18 and 88%, with scores of 57% and 88% for the two children in the same age range as that reported here.   Similarly, Roberts *et al.* (2005), report an average PCC of 55% for their male children aged from 4 to 13 years, and Dodd and Thompson (2001) report 54.8% for children between 5 and 15 years.

In terms of accuracy of imitation, Laws and Bishop (2003) report around 80% of 1- and 2-syllable words as repeated accurately by children with DS between 10 and 19 years of age. This score is somewhat higher than the 61% reported here.   This discrepancy is perhaps because the earlier study did not include children who wore hearing aids, whereas 1 of our participants was a hearing aid wearer, and four others had mild hearing loss.   Alternatively the discrepancy may be due to the mode of presentation.   Laws and Bishop presented items for imitation live by the experimenter, whilst we used an audio recording, so visual cues were not available.   Knowland *et al.* (in prep) found a small but significant benefit for *nonword* repetition tasks presented audio-visually to typically developing children up to nine years of age.   Further research could investigate the effect on imitation of audio-only, as compared to audio-visual, presentation for the DS population, who might well be expected to show an even greater benefit for audio-visual conditions that would allow them to draw on their strong visual skills.

Inconsistency

There was no significant effect of condition on inconsistency.   However, numerically, the read

productions were less inconsistent (45.8%) than those produced in naming (62.7%) or imitation

(59.1%) conditions.

Inconsistency in the naming condition was similar to that reported by Dodd and Thompson

(2001), who obtained an inconsistency score of 67% for 15 children with DS aged from 5 to 15

years.   Whilst children with DS are known to present with inconsistent productions, it is

noteworthy here that some of this inconsistency related to improving accuracy over the course

of the three repetitions.

This is the first time, to our knowledge, that inconsistency data has been reported for words

read or imitated by children with Down's syndrome.   Although not significantly different from

other conditions, it is encouraging to see that, in the reading condition, the inconsistency

percentage was nearing 40%, which is considered to be a clinical cut-off for inconsistent speech

disorder (Dodd *et al*., 2002).

It is possible that testing with additional participants and a greater number of words would

more clearly determine if reading words can indeed help to reduce inconsistency.   However, it

would be important to assess any such stimuli for phonetic and phonological balance.   For

example, the 25-word Inconsistency Test (Burt *et al.*.1999), used by Dodd and Thompson

(2001), could be adapted for reading and imitation, in order to further extend this study.

Intelligibility

There was a small but statistically significant effect of condition on ratings of intelligibility.

Reading productions were rated as more intelligible than those produced in either the naming

or imitation condition.   This finding tallies with the finding of greater accuracy of reading as

measured by PCC.   It should be noted though that the effect of reading is small, and its

practical value will need to be assessed in future studies.   Unlike the findings for accuracy,

productions in the imitation condition were found to be less intelligible than those produced

during naming. This finding would seem to indicate that listeners were affected by aspects of

intelligibility not captured by PCC, such as stress patterns, and vowel productions, which should

be investigated in future work.

In general productions in all conditions were rated as reasonably intelligible, with a mean score

of 3.27-3.74, where 5 represented a completely intelligible production. This might seem

surprising given that children with DS are frequently described as having unintelligible speech

(e.g. Cleland *et al.*, 2010).   The relatively intelligible speech in this study is probably due, in

part, to the reasonable accuracy of the children's productions, as noted above.   Recall,

however, that ratings were only given for the first repetition of each word in each condition,

and that accuracy was lower for this first repetition than subsequently, at an average of 58%

PCC.

Another factor that might contribute to the relatively high ratings is the method we used to

assess intelligibility (see Miller 2013, for a useful review of different approaches). Note that

raters had to know the target of the word they were rating, as it was crucial for them to hear

the same words multiple times, from different children and conditions. However, knowing

the targets may have caused raters to overestimate the intelligibility of the productions they

heard; we know for example, that transcriptions are more likely to be similar to the target

when that target is known (Oller and Eilers, 1975). Future work might employ different

designs for assessing intelligibility (see Kent, Miolo and Bloedel, 1994, for a review of evaluation

procedures), such as asking listeners to simply write the word that they hear, or, as in Cleland *et

al.*(2010), circle the correct option from a number of similar alternatives. It may also be

instructive to compare ratings of single words to a more global rating of individual children's

Mechanisms underlying the effect of reading

In sum then this study's results provide some tentative support for claims that reading words

improves accuracy and intelligibility of speech production for children with DS, although there

was only a numerical reduction in inconsistency when reading. In order to make suggestions

for intervention, and further understand the underlying nature of speech production deficits for

children with DS, it is important to consider the mechanisms behind any facilitatory effect of

reading. In order to do identify these mechanisms, the routes by which words are read aloud

must be considered.

Many models of skilled reading (e.g. Coltheart *et al.*, 2001) suggest that there are at least two

distinct routes for reading words aloud. New or unfamiliar words are read via a sublexical

route, using, for example, grapheme-phoneme conversion.   On the other hand, familiar and irregular words are read as a whole, using a lexical route.   Competition between these routes can account for phenomena such as regularity effects.   However, the reading route or routes used by children with DS are currently unknown.   Dodd and Crosbie (2005) recommend teaching a whole word approach, focussing on the lexical route, in order to link visual orthographic input with a phonological output plan, and many educators teach using a whole word approach in order to capitalise on the strong visual skills of children with DS.   However, Lemon and Fuchs (2010) demonstrate that children with DS <u>do</u> rely on phonological awareness skills when learning to read, and suggest that a phonics based approach, which would rely on sublexical reading routes, could be beneficial.   The current study did not aim to investigate the different routes by which children with DS read words, but continuing to pursue the issue of reading routes will be crucial for a full understanding of if, how and why reading improves speech production.   Further testing to elucidate this issue might compare performance on regular and irregular words; if the facilitatory effect of reading occurs because of accessing a sublexical route, then regular words should benefit more from reading aloud than irregular words.   If this effect is linked to whole word reading, via a lexical route, then regular and irregular words should benefit equally.   In addition, further analysis of error patterns in different conditions would also add to our understanding of the speech production of children with DS, and potentially help to clarify whether difficulties are related to oral motor, motor programming or phonological deficits.

Conclusion

Clearly this study is only a first step in fully assessing the efficacy of reading for children with DS, as we did not conduct an intervention study, did not test generalisation to other words or connected speech contexts, and did not assess if any gains for reading were maintained over time.   In addition, the numbers of participants and words tested were small.   These are all areas for future work and further research is needed in order to support or refute the current study's findings.   Nevertheless, these findings are encouraging and support earlier discussions highlighting the benefits of reading for children with DS.   Future studies will be useful to confirm this finding, and to identify the mechanisms behind any facilitatory effect of reading.

Appendix A

Participant information, via parental report.

Participant	Age	Gender	Speech Impairment	Speech intelligibility rating	Hearing impairment	Visual Impairment	Languages Other than English	Education	Started to read at age
1	11 ;6	female	Verbal dyspraxia	3	Mild loss and hyperacusis	Astigmatism	None	Special school	4 yrs
2	11;2	male	Delayed speech	3	None	Unilateral visual weakness	French	Mainstream	7 yrs
3	11;6	female	Yes, not specified	4	None	Bilateral visual impairment	Urdu	Mainstream	8 yrs
4	12;8	male	Yes, not specified	3	Mild loss	Astigmatism	None	Special school	3 yrs
5	11;7	female	Yes, not specified	4	High frequency loss	Long sighted	Portuguese	Mainstream	5 yrs
6	12;0	female	Yes, not specified	3	Moderate loss, hearing aids	Long sighted	None	Special school	5 yrs
7	13;3	female	Slight stammer	3	Mild left loss	Astigmatism	None	Special school	4 yrs
8	14;0	female	Articulation difficulties	3	None	Yes, not specified	None	Mainstream	4 yrs

Appendix B

List of stimuli

1) Jam

2) Stick

3) Glove

4) Yellow

5) Watch

6) Hand

7) Ship

8) Feather

9) Rings

10) Bath

Appendix C

Individual Results

Individual results for accuracy (PCC)

Participant	Naming	Reading	Repetition
1	63.89	63.22	64.44
2	45.83	62.07	52.78
3	53.45	61.36	48.15
4	65.56	71.26	60.71
5	47.22	82.64	64.49
6	62.18	75.93	61.11
7	74.71	66.09	65.52
8	66.00	72.44	73.46

Individual results for inconsistency (%)

Participant	Naming	Reading	Repetition
1	70	60	33
2	50	40	78
3	56	50	86
4	63	67	60
5	100	14	50
6	44	40	63
7	56	50	44

Individual results for intelligibility (mean rating)

Participant	Naming	Reading	Repetition
1	4.27	4.02	3.70
2	3.47	3.58	3.06
3	3.09	2.78	2.80
4	4.02	3.65	3.11
5	3.62	4.11	3.94
6	3.83	3.57	2.98
7	3.79	3.99	3.87
8	3.04	3.87	2.76

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