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ABSTRACT

This study aimed to investigate the association between listeners' ratings of how much effort it took to understand the speech of young people with cerebral palsy and the percentage of words listeners actually understood. Thirty-one young people with dysarthria and cerebral palsy (16 males, 15 females; mean age 11y, SD 3) were audio recorded repeating single words and producing speech. Objective measures of intelligibility were calculated for multiple familiar and unfamiliar listeners using a forced choice paradigm for single words and verbatim orthographic transcriptions for connected speech. Listeners rated how much effort it took to understand speech in each condition using a five point ordinal ease of listening (EOL) scale. Agreement on EOL within rater groups was high (ICC > 0.71). An effect of listener was observed for familiar listeners, but not for unfamiliar listeners. EOL agreement between familiar and unfamiliar listeners was weak-moderate (ICC=0.46). EOL predicted percentage of speech actually understood by familiar and unfamiliar listeners ($r > 0.56$, $p < 0.001$ for all predictions). Strongest associations between EOL and intelligibility were observed for speakers with mild and profound impairments. The findings of this study demonstrate that listeners can judge how well they have understood dysarthric speech. EOL is associated with listener familiarity, speech task and speech impairment severity. EOL is appropriate for use in clinical practice as a measure of communication activity.

Around 40% of children with cerebral palsy have motor disorders that affect their oromotor function, with dysarthria observed in around 36% (Parkes, Hill, Platt, & Donnelly, 2010). For children with dysarthria a primary aim of speech-language pathology is to maximise their speech intelligibility, with the end goal of facilitating children's access to and inclusion in family, educational and social activities (Taylor-Goh, 2005). A recent systematic review showed no conclusive evidence for interventions developed to improve speech intelligibility; but early phase studies of interventions developed from a theoretical understanding of dysarthria, which focus on respiratory support, speech rate and loudness, may be of benefit (Pennington, Miller, & Robson, 2009). These types of interventions continue to be investigated (Fox & Boliek, 2012; Pennington et al., 2013). Sensory-motor therapies, such as PROMPT are also now being evaluated (Ward, Strauss & Leitão, 2013; Ward, Leitão & Strauss, 2014). However, the current lack of conclusive evidence on the effectiveness of speech-language pathology interventions has led to wide-ranging interventions being employed (Watson & Pennington, submitted).

Whichever intervention is employed to improve children's speech intelligibility we need reliable and valid outcome measures with which to evaluate treatment success. Objective and subjective measures of intelligibility have been developed. Objective measures are the gold standard, showing both validity and reliability (Hustad, 2006, 2007; Hustad, Schueler, Schultz, & Duhadway, 2012; Miller, 2013; Pennington, Miller, Robson, & Steen, 2010; Pennington et al., 2013). They comprise calculations of the percentage of words that are understood correctly, achieved by asking listeners to transcribe the words they hear or by using a forced choice paradigm in which listeners select the word they have heard from a list (Hustad, 2006; Kent, Miolo, & Bloedel, 1994). However, objective measures are often labour intensive, requiring the recording of speech samples, the use of predesigned lists of words or sentences for forced choice paradigms, commitment of others to act as listeners and the checking of responses to calculate intelligibility. The resources required mean that objective measures of intelligibility are sometimes difficult to employ in busy clinical practice.

Subjective measures of intelligibility include: judgements of the percentage intelligibility of speech, whereby listeners estimate the percentage of words they have understood; visual analogue scales, in which raters mark the extent to which they think a person's speech is intelligible on a line of defined length with anchors representing completely unintelligible to completely intelligible; and direct magnitude estimation (DME), in which raters hear multiple recordings and rank the intelligibility of each recording against the others (Kent et al., 1994; Zraick & Liss, 2000). Visual analogue scales and DME have not, to our knowledge, been used in research into dysarthria in cerebral palsy. Estimated percentage intelligibility has been used (Hustad, 2007, 2008), is quick to perform and has been shown to be associated with objective percentage intelligibility (Hustad, 2006; Yorkston & Beukelman, 1978). Hustad observed that the relationship between perceptual and objective measures was affected by the severity of the speaker's speech impairment and by speech task. However, a direct relationship between the two measures was not achieved; some listeners overestimated and some underestimated intelligibility when compared with objective measures (Hustad, 2006).

Listener influences on intelligibility have received little attention in dysarthria research with children with cerebral palsy; although with better understanding of both speaker and listener impacts it may be possible to develop valid, reliable perceptual measures that could be used in clinical practice. The fact that associations between perceived and objective measures have been observed suggests that there are patterns in rater behaviour. One key question in this exploration concerns listeners' awareness of how well they understand dysarthric speech. Hustad (2007) began to examine this issue for listeners of people with cerebral palsy. She asked listeners to rate their confidence in their objective ratings of speech derived from transcriptions of recorded speech on a seven point scale (1 = not at all confident to 7 = very confident). Only weak to moderate associations were observed between confidence ratings and objective percentage intelligibility, suggesting that the listeners were not accurate judges of their understanding of dysarthric speech. Weak correlations may be due, in part, to the numerical severity rating scale used to measure confidence.

Stronger correlations may be achieved if raters are provided with descriptions to be used as rating anchor points in the measure.

A further listener aspect impacting on intelligibility is the amount of effort listeners need to expend in understanding the degraded speech signal, that is, the ease with which speech is understood (Laures & Weismer, 1999; Tjaden & Wilding, 2005). Whitehill and Wong investigated listeners' perceived effort in understanding speech of speakers with dysarthria associated with a range of neurological disorders, using visual analogue scales, and observed a strong correlation between effort and intelligibility (Whitehill, 2006). Listeners in a study by Miller and colleagues used a five point ordinal scale to rate the ease with which they could understand dysarthric speech of people with Parkinson's (Miller et al., 2007) e.g., 1 = No problem, easy to decide what the word/phrase was, and high levels of rater agreement were observed and strong correlations with objective measures of intelligibility. Although the dysarthria of people with Parkinson's will vary from that of young people with cerebral palsy, the wording of the scale developed by Miller et al. lends it to wider application including potential adoption in cerebral palsy research.

This study aimed to investigate further the relationship between listeners' actual understanding of the dysarthric speech of young people with cerebral palsy, measured by objective percentage intelligibility, and their perceptions of the effort exerted and the extent of their understanding, measured using an ordinal scale with anchored rating points. Objective intelligibility has been found to vary according to familiarity with the speaker and speech task (Hustad, 2007; Pennington & Miller, 2007; Pennington et al., 2010; Pennington et al., 2013). In this study we tested the association between perceived ease of understanding and actual understanding of listeners who were familiar with the speakers with cerebral palsy and listeners who had no experience of interacting with people who have speech disorders in single word and connected speech tasks. Our specific research questions were: 1. Is the ordinal scale of ease of listening developed by Miller et al. (2007) reliable when used with recordings of people with cerebral palsy? 2. Do familiar listeners report greater ease of understanding than unfamiliar listeners? 3. Does subjective rating of ease of

understanding predict objective percentage intelligibility for familiar and unfamiliar listeners? 4.

Does ease of understanding predict objective percentage intelligibility in single words and connected speech? 5. Is there an effect of severity of speech disorder on the association between ease of understanding and objective percentage intelligibility?

METHOD

Participants

Children receiving intervention

Thirty-one children with CP and dysarthria (sixteen males, fifteen females, age range 5-18y; mean 11y, SD 3) were recruited in the north of England via local speech-language pathologists, for previously reported studies (Pennington et al., 2010; Pennington et al., 2013). Children were eligible for the study if they had a diagnosis of CP, were aged of 5 to 18 years, and had dysarthria that was judged as moderate to severe by their local speech-language pathologists. Sex was not an issue for recruitment as there is no evidence that changes in voice production in puberty differs for adolescents with or without CP. Exclusion criteria were: bilateral hearing loss greater than 50 dB (which would affect ability to hear speech contrasts); severe visual impairments not correctable with spectacles (which would affect ability to see test material); and profound cognitive impairments or difficulties that would affect ability to follow the study instructions. Seventeen children had spastic type motor disorders, six had dyskinesia, four had mixed type motor disorder (spastic and dyskinetic), three had Worster-Drought syndrome (Clark, Chong, Cox, & Neville, 2009; Clark, Harris, Jolleff, Price, & Neville, 2009), and one had ataxia. Gross Motor Function Classification System (Palisano, Rosenbaum, Bartlett, & Livingston, 2007) levels ranged from I to V (I = 1; II = 12; III = 4, IV = 11; V = 3). Seven children were judged by their paediatricians to have IQ scores above 85 and fourteen were judged to have IQ scores 50-85, using the Surveillance of Cerebral Palsy in Europe

four level scale (Cans et al., 2007). In conversation children usually spoke in phrases (mean length of utterance in words 6.35; SD 2.45).

Listeners

To rate speech intelligibility, adults listened to recordings of children's speech. 270 adults with no experience of people with CP or disordered speech participated as unfamiliar listeners. Unfamiliar listeners were aged 18 – 60 years and recruited through business corporations in the North East of England. Three members of school staff who knew the participant children well were recruited as familiar listeners for each individual child aged 5-11 years (Pennington et al., 2013). Familiar listeners were not recruited to rate ease of listening for the older children. All listeners reported that they had no hearing difficulty, such as having to turn up the volume of their television or their families members reporting that they had misheard conversational speech.

Measures

The Children's Speech Intelligibility Measure (Wilcox & Morris, 1999) was used to elicit single word speech. The assessment comprises 200 lists of 50 single words, and in the test the children repeat each of the 50 words on the list as spoken by the tester. Different word lists were randomly allocated to each child at each recording. Intelligibility of connected speech was measured from children's answers to five simple questions, five repeated phrases, or descriptions of pictures up to one minute long. Children's answers were transcribed orthographically by the researcher and checked for accuracy with the child. Listeners were asked to give an ease of listening rating, based on how easy it was to recognise what the children were saying: 1 = No problem, easy to decide what the word/phrase was; 2 = Yes, I could tell what it was but I had to listen carefully; 3 = I just about recognised it with a bit of a guess; 4 = Complete guess, just going by vaguely recognised bits; 5 = Totally unrecognisable.

Procedure

This study combines separately collected but compatible data from two studies of an intervention for children with cerebral palsy and dysarthria which focussed on breath support, phonation and rate control. One study involved young people aged 12 to 18 years (Pennington et al., 2010), the second study was a replication involving younger children, aged 5 to 11 years (Pennington et al., 2013). Favourable opinion was obtained for the original studies from UK National Health Service research ethics committees. Children's guardians provided written consent for them to participate, and children gave written or verbal consent. Children's speech was digitally recorded (48kHz sampling rate; 16 bit quantisation) using EDIROL R9 (younger children in Pennington et al., 2013) or EDIROL R1 (older children in Pennington et al., 2009) recorders, and head-mounted microphones.

We recorded each child's speech on two separate days at four or five time points. We recorded older children who took part in the study reported in 2010 at six and one week before intervention and one and six weeks following its completion. We recorded the younger children in the 2013 study at the same time points and at twelve weeks post intervention. Thus, for older children we collected eight recordings and for younger children we collected ten recordings. We transferred all recordings to iTunes software (Apple Inc., Luxembourg). We randomly allocated recordings to each unfamiliar listener, so that they heard three recordings, each from different child. Each recording was heard by three different unfamiliar listeners. For familiar listeners we randomly selected one of the two recordings from each time point. Each familiar listener heard all of the selected recordings for the children they knew, but the order of presentation was randomised for each listener. We also randomised the order of presentation of single words and connected speech for each recording and for each listener.

Listening sessions were individual and took place at the listeners' places of work. Listeners were blind to the time points they were rating. They heard each recording only once, over loud speakers (Creative Inspire T12s) placed one metre away from them, without loudness adjustment.

In the single word condition, listeners selected the word they heard from a written list of twelve phonetically similar words. In the connected speech condition, listeners heard a phrase and orthographically transcribed the words they had heard. After each set of items (50 single words in the CSIM or connected speech samples), listeners rated the overall ease of listening out of five (1 = no problem, 5 = totally unrecognisable). Percentage speech intelligibility was calculated by dividing the number of words heard correctly by the number of words in the recording.

Statistical Analysis

Intelligibility scores took the form of the percentage of the words correctly understood by each listener. Previous studies (Pennington et al., 2010; Pennington et al., 2013) have already established inter-rater reliability for intelligibility for these data. The Ease of Listening (EOL) scale is a subjective ordinal measure. Inter-rater reliability had not been established for use with recordings of people with cerebral palsy, so intraclass correlation coefficients (ICC) were calculated for familiar and unfamiliar listeners. For unfamiliar listeners, ICCs were calculated using one-way random effects models. For familiar listeners, two-way mixed models (speech samples by raters) were used to calculate ICC, as the same raters judged each recording.

Linear regressions were used to test how well ease of listening score predicts intelligibility for familiar and unfamiliar listeners on the single word and connected speech items. Since intelligibility for each measure was not normally distributed, the regressions were repeated using squared ease of listening scores. Analysis was undertaken using SPSS for Windows, version 19 (SPSS Inc., Chicago, IL, USA). Intelligibility of speech to unfamiliar listeners was used to derive classifications of impairment severity following Hustad (Hustad, 2007). Mean intelligibility of 75% or above was classified as mild impairment (n = 47 recordings); 50- 74% intelligibility was classified as moderate impairment (n = 68); 25-49% was classified as severe impairment (n = 56) and less than 25% intelligibility was classed as profound impairment (n = 89)). An interaction between impairment

and ease of listening was fitted to the regression model to examine whether the strength of association between intelligibility and ease of listening depends upon the level of impairment.

RESULTS

Ease Of Listening inter-rater reliability

Inter-rater reliability mean ICC of EOL for familiar listeners hearing single words was 0.81, with a 95% confidence interval (95% CI) of 0.71 to 0.88. For connected speech inter-rater reliability mean ICC was 0.80 (95% CI 0.70 to 0.88). For unfamiliar listeners single word EOL inter-rater reliability ICC was 0.71 (95% CI 0.64 to 0.76). For connected speech, unfamiliar listener inter-rater reliability ICC was 0.81 (95% CI 0.76 to 0.85).

Effect of listener familiarity on ease of understanding

Agreement between familiar and unfamiliar listeners on EOL was moderate for single words (ICC 0.63; 95% CI 0.51-0.72). Agreement between listener groups on ease of listening to connected speech was also moderate (ICC 0.62; 95% CI 0.51-0.72). Paired t tests revealed that familiar listeners rated connected speech as easier to understand than unfamiliar listeners ($t(192) = 4.01, p < 0.001$). No difference was observed between familiar and unfamiliar listeners' EOL ratings of single word speech ($t(204) = -0.22, p = 0.08$).

Intelligibility and Ease of Listening

The intelligibility data initially appeared non-normal using Kolmogorov-Smirnov tests, so R-squared transformations were performed; however, since these were not successful in making the data normal, the untransformed data were used in the regression analyses. Boxplots showing the range of intelligibility scores associated with each EOL rating by familiar and unfamiliar listeners for single word and connected speech are shown in figure 1.

Insert figure 1 here

Familiar listeners' and unfamiliar listeners' ratings of EOL significantly predicted intelligibility for single words and connected speech and explained 31 – 63% of the variance in percentage speech intelligibility (table 1). For single words a reduction of one unit on the EOL scales was associated with an approximate decrease in intelligibility of 15% for familiar listeners and 16% for unfamiliar listeners. For connected speech a deterioration of 1 unit on the EOL scales was associated with an approximate decrease in intelligibility of 22% for familiar listeners and 23% for unfamiliar listeners.

insert table 1 here

Effect of impairment severity

Approximately 85% of the variance in percentage speech intelligibility was explained by familiar listeners' and unfamiliar listeners' ratings of EOL and impairment severity (table 2). There was no effect of interaction between EOL and severity in the prediction of single word or connected speech for either familiar or unfamiliar listeners. For familiar listeners only impairment severity predicted single word intelligibility. 'Severity and EOL predicted connected speech intelligibility for familiar listeners and single word and connected speech intelligibility for unfamiliar listeners.' Regression results are shown in table 2. Percentage distribution of ease of listening scores in the four severity groups are shown for single word and connected speech in figure 2.

Insert table 2 here

Insert figure 2 2

Discussion

The aim of this study was to investigate the association between listeners' perceptions of the ease with which they understand the speech of young people with dysarthria and cerebral palsy (the ease of listening) and the amount of speech they actually understand. We observed that the five point ordinal ease of listening scale developed by Miller and colleagues (2007) was reliable when applied with the speech of young people with dysarthria and cerebral palsy. Like previous research with other groups of dysarthric speakers (Miller et al., 2007; Whitehill, 2006), we found strong associations between perceived ease of listening and objective measures of intelligibility, with listeners understanding fewer words in recordings in which they expended greater listening effort. Our results also provide additional information on the effects of listener familiarity, speech type and speech impairment on ease of listening and its association with intelligibility.

Familiar listeners reported greater ease in understanding participants' connected speech than unfamiliar listeners, but a difference between listener groups was not observed for perceived ease of listening to single word speech. Our previous research showed that familiar listeners had understood more words spoken than unfamiliar listeners, with a greater difference observed between listener groups in connected speech than single word speech (Pennington et al., 2010; Pennington et al., 2013). Familiar listeners' greater ease of listening and actual understanding of connected speech is most likely attributable to the wider knowledge they can bring to bear in top down speech processing (Borrie, McAuliffe, & Liss, 2012; Borrie, McAuliffe, Liss, et al., 2012; D'Innocenzo, Tjaden, & Greenman, 2006; Flipsen, 1995; Kent & Read, 1992). In connected speech listeners use their grammatical knowledge to help decode the speech they hear (Kent et al., 1994). If listeners know the speaker they will be familiarised with frequently used phrases and speech mannerisms and use this information in auditory processing. Recognition of chunks or phrases (e.g., "well now", "you know", "I mean"), even if said only once or twice in a sixty second segment of connected speech, would be sufficient to boost the percentage of intelligible words correct and their

ease of listening. Listeners who know the speakers may also be able to recognise familiar articulatory errors and melodic intonation patterns and use these to decode words and phrases spoken. Although Whitehill and Wong (2006) found that segmental errors were more strongly correlated with listener effort than suprasegmental impairments, prosodic impairment has been correlated with intelligibility (Patel and Hustad et al. 2012) and could be playing a part in our results. Top down processing is not so readily available, and prosodic impairment will have little impact, in single word recognition tasks. Many of the words in the CSIM do not appear with high frequency in conversational speech. Auditory matches in the memories of familiar listeners would not be readily available and thus, knowledge of the speaker may provide limited advantage when listening to unconnected single words.

Stronger predictions of intelligibility by EOL were observed for unfamiliar listeners than familiar listeners, possibly because unfamiliar listeners were all hearing disordered speech for the first time and were therefore more similar as a group than familiar listeners, who had varying levels of familiarity with the speakers and with other young people with speech disorders in their professional roles. Higher levels of agreement on objective intelligibility had also been observed between Unfamiliar than between familiar listeners (Pennington et al., 2010; Pennington et al., 2013). Furthermore, because of their knowledge of the speakers, familiar listeners may also have had more investment in the listening tasks and some may have been unwilling to acknowledge how hard they needed to listen to the speakers. However, this speculation cannot be tested in the current study.

Similar to Hustad's observation of association between confidence in intelligibility rating and actual intelligibility for narrative speech (Hustad, 2007), we found that objective intelligibility was more strongly predicted by EOL in connected speech than single words for both Familiar and unfamiliar listeners. It is possible that the ease of listening is aided by the availability of both top down and bottom up processing. However, a limitation of our study is that we used different

methods to calculate connected speech and single word intelligibility. Connected speech was transcribed orthographically, whereas single words were selected from lists of phonetically similar foils. The foils may have interfered with EOL judgements. Furthermore, EOL ratings were made at the end of the intelligibility task. Connected speech took considerably less time to hear and transcribe than the fifty single words took to be heard and selected from their foils. Listeners may have found it difficult to provide an overall judgement on EOL for the fifty unconnected single words, many of which they may not have recalled when making their judgement. Recall would have been further aided in connected speech task by the cohesive nature of the speech sample, with words being encoded into a logical and more memorable unit than fifty unconnected single words (Baddeley, 1983).

In all conditions strongest prediction of intelligibility was observed for EOL rating of 1. Listeners who rated speech as being easy to understand usually perceived most of the speech correctly. Mildly impaired speech may be easy for all listeners to understand in all speech conditions. Speakers with low levels of intelligibility were most likely to be rated as being difficult to understand, with EOL rating of 4 or 5. However, there were outliers who reported severe difficulty in listening but who actually perceived large proportions of the speech correctly, especially in connected speech. It is possible that the contextual/linguistic information available in connected speech is increasing intelligibility for more severely impaired speakers but that listeners are still not confident in their use of this information. Weakest predictions were observed for EOL ratings of 3, where listeners acknowledged that they were guessing what the speaker had said (figures 1-4). EOL ratings of 3 can be found across the severity groups (figures 5-8), possibly explaining the lack of interaction between EOL and impairment severity given the clear reduction in frequency of EOL rating of 1 and 2 and increase in EOL rating of 4 and 5 with impairment severity. Wide variation in intelligibility for EOL rating of 3 may be due in part to the wording of the classification (I just about recognised it with a bit of a guess), as the amount of speech guessed is not stipulated. Further research is needed to

examine whether patterns in listener or speaker characteristics may also account for such wide variation at EOL rating 3, and why some guesses are correct and others not.

This study used audio recordings of repeated single words and connected speech to test the association between ease of listening and objective measures of intelligibility. Our connected speech samples comprised picture description, repetition of familiar phrases and answers to questions. Repetitions may not be processed in the same way as spontaneous speech; for instance, they will not involve word selection but they will rely on auditory working memory. We did not test the difference between the types of speech in our samples, but we envisage that any differences observed would be minimal as the repetitions comprised only a small subset of our samples. Nevertheless it should be borne in mind that our findings may not translate to audio recordings of other types of speech, such as conversation, or to video recordings, where further but perhaps competing information is available to the listener. Further research is needed to test the relationship between ease of listening and intelligibility with different speech stimuli.

Conclusion

This study has shown a clear relationship between listeners' perceptions of the ease with which they understand the speech of young people with dysarthria and cerebral palsy and the amount of speech they actually understand. This relationship exists for familiar and unfamiliar listeners and when listening to single word and connected speech. We conclude that EOL is appropriate for use in clinical practice as a measure of communication activity, adding a conversation partner perspective on communication success. However, the amount of variation in intelligibility observed for EOL ratings in the middle of the scale, when listeners report they are guessing what speakers have said, mean that we cannot use ease of listening as a proxy for objective intelligibility measures. Further research is needed to investigate if specific features of speakers or listeners are associated with correct or incorrect guesses.

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Table 1: Regression analyses for prediction of speech intelligibility by Ease of Listening score for familiar and unfamiliar listeners

		r^2	B	SE	β
Familiar listeners	Single Word	0.31	-15.02	1.53	-0.56*
	Connected Speech	0.47	-21.59	1.60	-0.69*
Unfamiliar listeners	Single Word	0.46	-16.79	0.65	-0.68*
	Connected Speech	0.63	-22.67	0.62	-0.80*

* $p < 0.001$

Table 2: Regression analyses for prediction of speech intelligibility by Ease of Listening score and speech impairment severity for familiar and unfamiliar listeners

		Full model		B	SE	β
		r^2				
Familiar listeners	Single Word	0.86	EOL	1.33	1.77	0.05
			Severity	-21.13	2.23	-0.84**
			EOLxSeverity	-0.69	0.70	-0.13
	Connected Speech	0.86	EOL	-3.97	1.98	-0.13*
			Severity	-22.76	2.74	-0.87**
			EOLxSeverity	0.20	0.81	0.04
Unfamiliar listeners	Single Word	0.85	EOL	-3.60	1.07	-0.15**
			Severity	-19.71	1.12	-0.81**
			EOLxSeverity	-0.43	0.37	-0.01
	Connected Speech	0.86	EOL	-6.87	1.38	-0.67**
			Severity	-18.73	1.23	-0.84**
			EOLxSeverity	-0.34	0.41	-0.07

EOL= Ease of listening

* $p < 0.05$, ** $p < 0.01$

Figure 1 Box plots showing distribution of single word and connected speech intelligibility by EOL rating for familiar and unfamiliar listeners

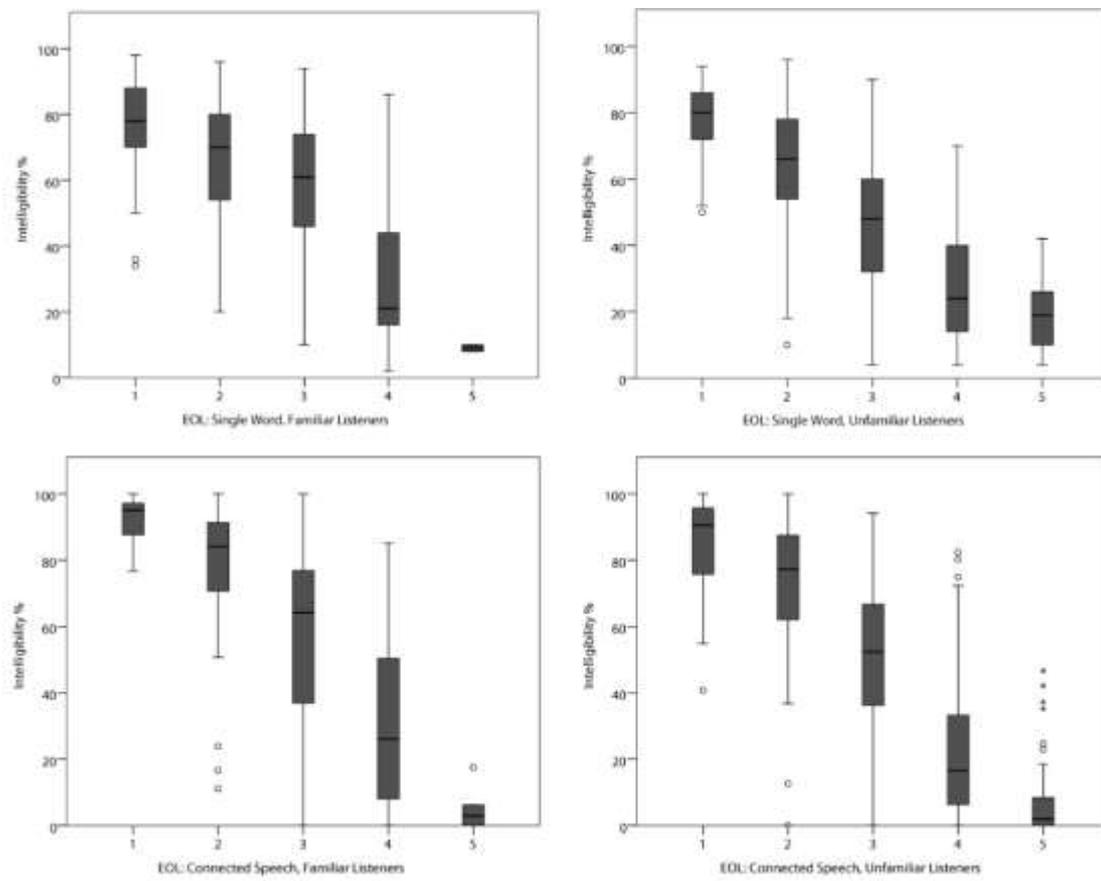


Figure 2 Percentage distribution of EOL ratings for the four speaker severity groups under single word and connected speech conditions

