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Executive functioning and verbal fluency in children with language difficulties

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Running head: Executive functioning, verbal fluency and language difficulties

Abstract

This study provided a detailed analysis of verbal fluency in children with language difficulties, and examined the relative contributions of executive functioning (executive-loaded working memory, switching, inhibition) and language ability to verbal fluency performance. Semantic and phonemic fluency, language, and executive functioning tasks were completed by 41 children with specific language impairment (SLI) and 88 children with typical development. Children with SLI showed difficulties with most aspects of verbal fluency (rates of output, errors, switching) relative to typical children. Language ability predicted nearly every aspect of phonemic fluency performance and some aspects of semantic fluency performance. The relationships between verbal fluency task, but relationships with executive-loaded working memory and switching were absent. Educationally, these results emphasise the underlying importance of language abilities in generation tasks like verbal fluency, but point to the importance of inhibition skills for error monitoring. Interventions to improve search and generation abilities have the potential to offer broader benefits in the classroom for children with language difficulties.

Keywords: language difficulties, verbal fluency, executive functioning, specific language impairment

1. Introduction

1.1 Executive functioning and children with language difficulties

Executive functioning (EF) describes a constellation of related abilities involved in high-level, goal-directed behaviour/self-regulation (Miyake & Friedman, 2012) to enable negotiation of complex and changing circumstances in the absence of automatic or fixed ways of responding (Diamond, 2013). The most influential model of EF in adults (Miyake et al., 2000) identifies three components: executiveloaded working memory (ELWM: the ability to process and store information concurrently); switching (the ability to rapidly and flexibly change cognitive set); and inhibition (the ability to suppress readily available responses/stimuli). Identifying these areas of EF in children has been broadly successful, although some studies report two factors (Fisk & Sharp, 2004; Huizinga, Dolan, & van der Molen, 2006; Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003; St Clair-Thompson & Gathercole, 2006; van der Ven, Kroesbergen, Boom, & Leseman, 2013), and recent conceptualisations have suggested inhibition might be part of a 'common' EF factor (Miyake & Friedman, 2012).

Children with developmental disorders show EF difficulties (e.g., ADHD, ASD: Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2009; Geurts, Verte, Oosterlaan, Roeyers, & Sergeant, 2004), and the current study focused on children with specific language impairment (SLI). Although SLI is a controversial label (Bishop, 2014; Reilly, Tomblin, et al., 2014), researchers and clinicians agree that a heterogeneous group of children with significant language difficulties compared to their peers can be recognised (Reilly, Bishop, & Tomblin, 2014). The current study defined SLI as a developmental disorder involving delayed receptive and/or expressive language (phonology, vocabulary, grammar) in the absence of any obvious cause (Bishop & Norbury, 2008). Although exclusionary definitions can be controversial (Reilly, Tomblin, et al., 2014), it is thought that SLI affects 3-6% of schoolchildren (Hulme & Snowling, 2009). Research classifying subgroups of children with different types/combinations of

language difficulty has identified verbal sequential memory, speech production, lexical-semantic abilities and auditory conceptualisation factors (Van Weerdenburg, Verhoeven, & Van Balkom, 2006), but pinpointing stable subgroups over development is challenging (Conti-Ramsden & Botting, 1999; Reilly, Bishop, et al., 2014) and was, therefore, not attempted in the current study. We identified children diagnosed as having SLI and additionally checked that they also obtained poor scores on a standardised language test. The resulting sample is likely to have been heterogeneous in terms of language difficulties, comprising individuals with a range of expressive and receptive impairments. Although definitions of SLI no longer use 'cognitive referencing' (Bishop, 2014), we nevertheless ensured that the current sample of children with SLI had non-verbal IQ scores in the average range or above, so we could assess difficulties in verbal fluency without a potential confound of low IQ. A comparison sample included typical children with a similar range of chronological ages, but we included some younger typical children to reflect the lower 'language age range' of the SLI group. All comparison children were assessed to have no current language difficulties.

Children with SLI have difficulties with EF. Inhibition is impaired (Bishop & Norbury, 2005; Henry, Messer, & Nash, 2012; Im-Bolter, Johnson, & Pascual-Leone, 2006; Weyandt & Willis, 1994), and ELWM is also impaired in many children with SLI (see Montgomery, Magimairaj, & Finney, 2010 for a review; see also: Archibald & Gathercole, 2007; Ellis Weismer, Evans, & Hesketh, 1999; Henry et al., 2012; Im-Bolter et al., 2006; Marton, 2008; Marton & Schwartz, 2003; Montgomery, 2002). Current developmental conceptualisations of ELWM suggest increases in short-term memory (STM) and processing speed are implicated for typical children (Bayliss, Jarrold, Baddeley, & Leigh, 2005), and both STM and processing speed (general or specific to linguistic tasks) are indeed weak in children with SLI: (STM: Bishop, North, & Donlan, 1996; Dollaghan & Campbell, 1998; Edwards & Lahey, 1998; Gathercole & Baddeley, 1990; Hick, Botting, & Conti-Ramsden, 2005; processing speed: Leonard et al., 2007). Archibald and Gathercole (2007) argued persuasively that both phonological storage and EF are

impaired in children with SLI. The only major unimpaired EF area in children with SLI is switching (Dibbets, Bakker, & Jolles, 2006; Henry et al., 2012; Im-Bolter et al., 2006; Kiernan, Snow, Swisher, & Vance, 1997; Weyandt & Willis, 1994, but see Marton, 2008). The current study examined all three areas of EF to assess their relationships to verbal fluency in a mixed sample of children with and without SLI.

1.2 Verbal fluency and executive functioning

Verbal fluency tasks assess "strategic search and retrieval processes from the lexicon and semantic memory" (Sauzéon, Lestage, Raboutet, N'Kaoua, & Claverie, 2004). They require generation of as many words as possible within one minute according to simple rules that target sounds (phonemic fluency, items starting with particular letters such as "f", "a", "s") or semantic categories (semantic fluency, "animals" or "foods") (Troyer, 2000; Troyer, Moscovitch, & Winocur, 1997). These two tasks measure related processes in adults (Unsworth, Spillers, & Brewer, 2011) and are correlated in children (Matute, Rosselli, Ardila, & Morales, 2004; Riva, Nichelli, & Devoti, 2000). Successively generated items are often related to each other along task-relevant dimensions: e.g., phonemic fluency relationships via spelling- sound knowledge; or semantic fluency tasks are an important window into children's' lexical/semantic/phonemic networks and strategic search and retrieval processes (Sauzéon et al., 2004). As might be expected, verbal fluency abilities improve with age in typical children (Hurks et al., 2010; Kavé, 2006; Kavé, Kigel, & Kochva, 2008; Klenberg, Korkman, & Lahti-Nuuttila, 2001; Korkman, Kemp, & Kirk, 2001; Matute et al., 2004; Riva et al., 2000; Sauzéon et al., 2004).

Verbal fluency is often described as a measure of EF (Pennington & Ozonoff, 1996), and has been related to executive dysfunction after neurological damage (Henry & Crawford, 2004) because it requires goal-directed behaviours such as flexibility of thought, strategic planning, non-habitual responses and error-monitoring. Diamond (2013) suggested verbal fluency reflects one aspect of

cognitive flexibility. Contrastingly, verbal fluency has been regarded as a language measure and related to lexical access (semantic and phonological retrieval processes), language proficiency, vocabulary size (Luo, Luk, & Bialystok, 2010), and vocabulary knowledge (Prigatano & Gray, 2008; Ruff, Light, Parker, & Levin, 1997). Importantly, children with SLI (Weckerly, Wulfeck, & Reilly, 2001) and those with other language difficulties (dyslexia: Cohen, Morgan, Vaughn, Riccio, & Hall, 1999; deaf signers with SLI: Marshall, Rowley, Mason, Herman, & Morgan, 2013; word finding difficulties: Messer & Dockrell, 2013; Down Syndrome: Nash & Snowling, 2008) show difficulties with verbal fluency.

The current study had two aims. (1) To explore whether verbal fluency performance limitations in children with SLI can shed light on underlying difficulties. (2) To test current theoretical conceptualisations of the relative roles for EF and language ability in *predicting* verbal fluency performance. These two issues are reviewed below.

1.3 Aim 1: What does verbal fluency reveal about children with language difficulties?

Verbal fluency is a multifactorial task requiring a range of performance factors (Troyer et al., 1997). Generating items according to a rule taps the ability to search and retrieve relevant information from lexical/semantic memory (Kavé et al., 2008), particularly, the ability to retrieve phonological and semantic information (Marshall, 2014). It reflects monitoring of output for errors/rule violations (Unsworth et al., 2011), and provides information about the organisation of and access to semantic/phonemic networks (Nash & Snowling, 2008; Weckerley et al., 2001). These are highly relevant and generalizable skills/abilities in the classroom, involved in learning activities whereby organised, stored information on a range of topics (e.g., chemistry, history, physics, languages etc.) is accessed and retrieved according to relevant dimensions and monitored for accuracy/relevance.

These processes were investigated in detail. Total number of items generated in phonemic and semantic fluency tasks assessed ability to search for, retrieve and generate information from

semantic/phonemic networks ('total output'). Correct items ('valid output') within task rules may be more relevant to classroom learning, demanding refined searches, careful response selection and ongoing monitoring for accuracy. Further, to gain insight into accuracy monitoring, errors were assessed. Previous work (Weckerly et al., 2001) reported that children with SLI produced fewer responses, but showed no differences in numbers of overall errors, suggesting difficulties with search, retrieval and generation of items, not with monitoring failures. However, Weckerley et al.'s (2001) measure of total errors did not account for overall level of performance, so a *proportion* of errors measure was calculated here.

Verbal fluency tasks are powerful methods of assessing switching between clusters of related items (Troyer et al., 1997). Although EF switching abilities appear preserved in children with SLI (see earlier), in such tasks, categories are detected from visual cues (e.g., Wisconsin Card Sorting Task). Verbal fluency switching requires *self-generation* of new sub-categories with no cues. Children with SLI may be less able than typical children to self-generate relevant categories and to switch between 'exhausted' sub-categories of target items. This could reflect less extensive or well-organised semantic/phonemic networks, limited semantic/phonemic/lexical knowledge (having fewer subordinate categories available) (Nash & Snowling, 2008), and/or reduced efficiency in accessing stored knowledge (Marshall, 2014).

Relationships between adjacent items generated are often examined for 'clustering' (semantic relations, "dog, cat, hamster"; or phonemic relations, "fast, fat, far") (Troyer et al., 1997), giving insight into the extent, organisation, and accessibility of lexical/semantic and phonemic networks (Nash & Snowling, 2008; Troyer, 2000; Troyer et al., 1997). Smaller clusters of related items in children with SLI than typical children would suggest reduced ability to perform exhaustive searches, and/or smaller semantic/phonemic networks of related items. Both factors might impact negatively on learning and

organising new information efficiently in semantic memory to achieve successful and rapid access. Evidence for reduced cluster sizes in children with SLI could support the use of interventions to elaborate and enhance semantic representations (for a review see Cirrin & Gillam, 2008) and to impose clear structures when learning new information (e.g., mind maps). Although cluster size did not differ between children with SLI and typical comparisons in the Weckerly et al. (2001) study, further data will allow stronger conclusions about this important issue.

Automatic and controlled processing was assessed using response rates, as these typically decline over the one-minute output period. It has been suggested that the first 15 seconds of word generation reflect automatic processes for accessing common/prototypical/easily accessible items. Once readily accessible items have been exhausted (subsequent 45 seconds), effortful 'controlled' search processes are required, placing heavier loads on EF (Hurks et al., 2006). Children with SLI may resemble typical children during the first 15 seconds of output (automatic processing), whereas the controlled processing phase may reveal group differences. Alternatively, children with SLI may show weaker performance throughout the output period due to slower processing speeds (Leonard et al., 2007) or poorly executed search processes. Absolute and relative changes in performance during the response output period will provide new and important data with educational implications. If automatic processing is preserved in children with SLI, this would suggest that consolidating the learning of key basic concepts to achieve 'automaticity' might be beneficial.

Therefore, children with SLI were assessed for difficulties with searching semantic/lexical memory and generating responses, monitoring output for accuracy, switching between and exhaustively searching clusters of related items, linking related items together, and controlled versus automatic generation processes. The general prediction (Hypothesis 1) was that children with SLI would show difficulties with many if not all of these measures. The current mixed sample of children with SLI is likely

to include children with difficulties with phonology, grammar and/or vocabulary, which may differentially affect phonemic and semantic fluency, but as the sample was not stratified, predictions were formulated at the level of 'group performance'.

1.4 Aim 2: Is verbal fluency performance related to executive functioning and/or language ability?

According to neuropsychological evidence reviewed by Troyer et al. (1997) (lesion studies, fMRI, PET), executive control processes are crucial in verbal fluency tasks: ELWM to monitor output for relevance and retain the 'rules' of the task; and inhibition to control the repetition of responses (Unsworth et al., 2011). Switching, or cognitive flexibility, may be required to switch between clusters of related items (Hurks et al., 2010; Troyer et al., 1997). EF is likely to be more relevant for phonemic than semantic fluency tasks (Riva et al., 2000; Sauzéon et al., 2004; Troyer et al., 1997). This is because phonemic fluency is harder than semantic fluency (e.g., Hurks et al., 2006; Troyer, 2000), requiring effortful searching through larger numbers of subcategories (Hurks et al., 2006) that rely on unusual and demanding orthographic criteria, rather than the organisational structure of everyday life (Henry & Crawford, 2004).

A further predictor of verbal fluency was suggested by Luo et al. (2010), namely 'language ability' (assessed with a vocabulary measure). Luo et al.'s work was premised on findings that bilingualism from an early age conveys *advantages* on EF (via extended practice with switching and resolving conflicts between languages), yet *disadvantages* on highly constrained verbal tasks such as vocabulary (Bialystok, 2010, 2011). By dissociating these factors in bilinguals, Luo et al. showed that both were important for verbal fluency performance, specifically in relation to phonemic fluency, and made the following points: (1) EF is a more important predictive factor for phonemic than semantic fluency (more difficult search processes are required); (2) language ability is a more important predictive factor for semantic than phonemic fluency (larger/more integrated semantic/lexical knowledge); (3) EF

is increasingly required as the trial progresses (once easily accessed items have been exhausted and demands on strategic search processes increase); and (4) language ability is relevant throughout a response trial.

Following this theoretical conceptualisation, we derived further predictions. Hypothesis 2 was that EF should be a stronger predictor of phonemic than semantic fluency, whereas language ability should be a stronger predictor of semantic than phonemic fluency. Hypothesis 3 was that EF should show stronger relationships with performance in the later stages of each response trial, whereas language ability should remain a predictor of performance throughout the response trial. Language ability was assessed using raw scores from two verbal tasks in a standardised test battery. EF was assessed using verbal measures of ELWM, inhibition and switching, in line with current models (Miyake et al., 2000). The sample included children with large ranges of abilities in both language and EF, so relationships between the variables should be more likely than in a homogeneous sample.

2. <u>Method</u>

2.1 Participants

Children with SLI (8yrs1m to 14yrs1m) were recruited from mainstream schools and specialised language units in Greater London, via letters to Headteachers and/or Special Needs Co-ordinators asking them to identify children with language difficulties. In approximately half of the schools that took part, children without language difficulties (6yrs0m to 14yrs8m) were recruited from the same classes to ensure that the groups were broadly similar in terms of socio-economic background. We also recruited some younger typical children from schools in similar areas to reflect the full range of approximate language ages of those in the SLI group. The 129 participants came from 22 schools and specialist language units/classes and, very occasionally, via direct contact with parents/guardians. The final samples comprised typical children (n=88, mean (SD) age 117.7 (28.1) months) and children with SLI (n=41, mean (SD) age 138.4 (15.9) months). Age differed significantly between groups, t(122.12)=5.31, p<.001, so was controlled statistically in all analyses. Key background variables are presented in Table 1. A further 32 potential participants were excluded because they did not fit the study criteria outlined below (language difficulties in the milder range; non-verbal IQ in the borderline or intellectual disability range).

All participants in the SLI and typical groups spoke English as their first language and had nonverbal IQs in the average range [T-scores >40 on BAS-II Matrices (British Ability Scales-II, Elliott, Smith, & McCullouch, 1997), standardised to give mean T-score=50, SD=10]. There is disagreement concerning IQ cut-offs and SLI, but many researchers specify 'average' levels of non-verbal IQ (Bishop, 2014): this criterion was adopted in the current study to examine the role of language difficulties (in the absence of low IQ) on verbal fluency. BAS-II Matrices scores differed between groups [mean (SD) T-scores: SLI=54.6 (6.2); TD=57.4 (6.9)], t(127)=2.21, p<.05, so were statistically controlled in all analyses.

Children in the SLI group had formal diagnoses from appropriate health professionals according to standard clinical criteria and no diagnoses of hearing impairments, intellectual disability, or other developmental disorders (e.g., ADHD, ASD). Inclusion in the SLI group was dependent on additional screening we carried out: participants had at least three/four scaled scores at or below 1SD (≤7 where scaled score mean=10; SD=3) on subscales from the CELF-4-UK (Clinical Evaluation of Language Fundamentals-4-UK, Semel, Wiig, & Secord, 2006). This standardised measure is used in the UK by Speech and Language Therapists to assess and identify children with language difficulties; usual clinical cut-offs are around one standard deviation below the normative sample mean. We administered Recalling Sentences (reliability .90), Formulated Sentences (reliability .86), Word Classes-Receptive (reliability .86) and Word Classes-Expressive (reliability .83) to obtain a broad view of expressive/receptive language abilities in this heterogeneous sample. These tasks represent robust

measures and include the most sensitive CELF-4 subtests, as evidenced by intercorrelational, clinical validation and diagnostic studies, showing the test to be sensitive to variability in language difficulties. All participants in the typical group had scaled scores of 8 or higher on the four CELF-4-UK subscales.

The known gender imbalance in those with SLI favouring boys (Leonard, 1998) was reflected in the typical sample, which was larger than the SLI group to ensure it spanned the range of language/chronological ages of the children with SLI (see Table 1). This study did not utilise a group matching procedure, opting instead for the use of multiple regression analyses to control for key variables usually matched between groups (age, non-verbal IQ). This maintained the representativeness of the sample by avoiding exclusion of unmatched participants.

Table 1 about here

2.2 Study Variables

2.2.1 Verbal Fluency: This was assessed using relevant tasks from the Delis-Kaplan Executive Functioning System (Delis, Kaplan, & Kramer, 2001, D-KEFS). Children generated as many words as possible in one minute according to a set of rules. The words had to start with a particular letter (phonemic fluency: letters F, A, S); or belong to a particular semantic category (semantic fluency: 'animals', 'boys' names'). Repeated items and items outside the set were counted as errors. The following measures were calculated: total number of words generated inclusive of errors; total number of valid words (exclusive of errors); total number of errors; proportion of errors in relation to total number of words generated; and numbers of valid words generated in each of four 15-second response quartiles.

Two further measures were calculated, based on the relatedness between successive items generated in the phonemic and semantic fluency tasks, according to criteria set out by Troyer et al. (1997). For phonemic fluency, successively generated words beginning with the same first two letters were classified as belonging to the same cluster. In one or two rare cases, successive words with the same first two sounds were accepted if spelling patterns were not identical (skid, scam; sky, school). Words that rhymed (*sip*, *slip*) or that were identified by the child as homophones (*sea*, *see*; *sum*, *some*) were included in the same cluster, as were items that differed in a single vowel sound (*sin, son, sun*; feet, foot, fat). For calculating item relatedness in the semantic fluency tasks, we excluded the semantic category of 'boys' names' because it was impossible to assess semantic relatedness for individual children. Therefore, these measures reflect the category of animals. Groups of related animals were based mainly on Troyer et al., but we included some further groups of related animals generated by children in the sample. Sub-categories included sea creatures, pets (including sub-categories of dogs), farm animals, African mammals, insects, small reptiles, big cats, Australian animals, British mammals, American mammals, birds, primates, snakes, large reptiles. Single words and groups of words counted as separate clusters. In many previous studies, mean size of cluster has been counted only from the second word within a cluster (Mayr, 2002), but this approach was considered potentially misleading because many children had more single word 'clusters' than multi-word clusters. For each child, the following scores were calculated: mean number of switches between clusters (single words and groups) and mean number of words per cluster. Reliability of scoring was assessed for 15% of the sample rated by a second coder. The agreement between coders was r = .998 for letter fluency and r = .960 for animal fluency.

Summary descriptive data on all of the fluency variables for each group are given in Table 2: note that these scores are not directly comparable because the groups were not matched for age and IQ (these factors were controlled using multiple regressions). The five measures of verbal fluency (valid responses) were all significantly related to each other, with correlations of .49-.78 (mean r=.60).

2.2.2 Language ability: This was assessed using raw ability scores from the BAS-II Word Definitions (expressive vocabulary, reliability .89) and Verbal Similarities (identifying similarities across sets of three words, "How are spoon, fork and knife alike?", reliability .91). These subtests provided a well-established and highly relevant measure of language ability, tapping vocabulary knowledge, general semantic knowledge and expressive language (Elliott et al., 1997). Averaged raw ability scores were used rather than standardised scores to preserve maximum variability across age and ability (see Table 1). The SLI and TD groups differed significantly, as expected, on language ability [mean (SD) raw scores: SLI=100.8 (18.3); TD=111.5 (21.5)], *t*(127)=2.75, *p*<.01.

2.2.3 Executive-loaded working memory, inhibition, switching:

Executive-loaded working memory (ELWM). The task was Listening Recall (Working Memory Test Battery for Children, WMTB-C, Pickering & Gathercole, 2001). The Experimenter read a series of short sentences and the child judged whether each was true/false (processing), then recalled the final word from each sentence in correct serial order (storage). Trials commenced with list lengths of one item, and proceeded to longer lists, with six trials per list length, until fewer than 4/6 trials were correct. Total trials correct were scored, as this is more reliable than 'span' (Ferguson, Bowey, & Tilley, 2002). Testretest reliabilities of .38-.83 are reported for relevant ages (Pickering & Gathercole, 2001)

Inhibition. Verbal Inhibition, one of the sub-tests from the VIMI task (Henry et al., 2012), was employed. The Experimenter said either 'doll' or 'car' and the participant copied by repeating the same word (block 1). In block 2, the child inhibited this copying response: 'if I say doll, you say car; and if I say car, you say doll'. A second 'copy' and 'inhibit' block followed and each of the four blocks consisted of 20 trials. The entire sequence of copy/inhibit blocks was repeated in Part B with new stimuli ('bus', 'drum'). Combined errors across Parts A and B were used as the measure of inhibition. Cronbach's alpha, based on total error scores from Parts A and B was .727.

Switching. The Trail Making Test (D-KEFS) assessed switching. Children joined small circles containing letters and numbers alternately, in sequence (1-A-2-B-3-C through 16-P). Four control conditions assessed component skills. Most relevant were: Number Sequencing (connecting the numbers 1-16); and Letter Sequencing, (connecting the letters A-P). "Switching cost" was the time per item taken for combined letter/number switching, minus the sum of the time taken per item for the number and letter sequencing component skills. Test-retest reliabilities for measures contributing to "switching cost" are reported as: number sequencing (.77), letter sequencing (.57); letter/number switching (.20, Delis et al., 2001). Reliability for switching measures can be low, given they are difference scores; consequently, somewhat lower reliabilities may be inevitable (Miyake et al., 2000).

The project was granted ethical approval from the Research Ethics Committee, London South Bank University, and was discussed in detail with appropriate school staff before recruitment. Informed consent for participation was obtained in writing (telephone permission occasionally) from parents/guardians; children/students also gave their written consent and were told they could opt out at any time. The full testing protocol took place across 3-8 sessions, making up 3½ hours for the complete battery, usually at school but occasionally at the child's home. Test order was somewhat flexible to accommodate school timetables, but generally followed this protocol: BAS-II; CELF-4-UK; ELWM; verbal fluency; switching; inhibition. A number of other executive functioning tasks not reported here were also included (see Henry et al., 2012).

Table 2 about here

3. <u>Results</u>

For all regressions reported, key statistical checks (Durbin-Watson, tolerance/VIF statistics, Cook's/Mahalanobis distances, standardised DFbetas, leverage values, plots of standardised residuals/predicted standardised values, standardised residuals, partial plots) were carried out. Error variables were transformed using square root (total errors, inhibition errors) or arcsine (proportion errors) transformations, as these data were not normally distributed and initial checks indicated that individual cases may have had undue influence (Field, 2013). Alpha was set at p<.005 after Bonferroni corrections, based on 10 regressions per set of analyses, but tables include significance values at p<.05 for information.

3.1 Aim 1: What does verbal fluency performance reveal about children with language difficulties?

Group differences on all measures of verbal fluency were assessed to evaluate Hypothesis 1 (children with SLI would show difficulties with many if not all aspects of verbal fluency). Instead of attempting to match children in each group for age and non-verbal IQ, an alternative approach was taken to keep samples as representative as possible and avoid excluding unmatched children. Differences between the groups (SLI, typical development) in age and non-verbal IQ were controlled statistically using hierarchical multiple regression. For each verbal fluency variable, the regression controlled for age and non-verbal IQ at step one and introduced a dummy-coded group variable to assess group differences at step two. An alternative is to use analysis of covariance to control for age and non-verbal IQ, but this involves statistical assumptions not always merited by the data. Regression techniques are more robust.

Separate hierarchical regression analyses on the following dependent variables were carried out: *response output* (total, valid), *output over time* (patterns of performance over quartiles within the one-minute response period), *errors* (total errors, proportion errors), *switching* (number of switches between categories), and *cluster sizes*.

Tables 3 and 4 present key summary data. Each dependent variable is listed in Column 1. Column 2 shows total variance accounted for by the final model for each variable (and significance of overall model), and Column 3 gives the change in variance at step two (change in R^2), reflecting whether introducing the dummy-coded group variable accounted for further significant variance in the model (reflecting a group difference in performance). Columns 4 to 6 provide the standardised beta-values at step two of each regression model for the three predictor variables.

For semantic fluency (Table 3), the change in *R*² values at step two indicated significant group differences for the following measures: total responses, valid responses, performance in quartiles 1, 2, 3, and 4, total errors, proportion errors, and number of switches. Group accounted for 6-24% of the variance in performance on the fluency measures. Inspection of beta-values for the SLI-vs-typical comparison indicated that children with SLI produced significantly fewer responses (total, valid) than typical children, and showed reduced numbers of accurate responses in all four quartiles of the task. Children with SLI made more errors (total errors, proportion errors) and fewer switches within semantic categories. The only area in which children with SLI did not differ from typical children was words per cluster (this was also a non-significant regression model).

Table 4 shows the regressions for phonemic fluency. The change in R^2 values at step two indicated significant group differences for the following measures: total responses, valid responses, performance in quartiles 1, 2, 3, and 4, proportion errors, and number of switches. Group differences on words per cluster were marginally significant (p=.006). Group accounted for 6-33% of the variance in performance on the fluency measures. Inspection of beta-values for the SLI-vs-typical comparison indicated that children with SLI produced significantly fewer responses (total, valid) than typical children, and fewer responses across all quartiles of the task. Children with SLI produced a higher proportion of errors, made fewer switches within phonemic categories, and included marginally fewer words per cluster. There was one area in which children with SLI did not differ from typical children: they did not show an elevated absolute number of errors (see also Weckerly et al., 2001).

<u>Aim 1 summary</u>: for both semantic and phonemic fluency, children with SLI showed lower rates of overall response output (total, valid), lower performance on all four quartiles of the task, higher proportions of errors, and fewer switches compared to typical children. As children with SLI demonstrated wide ranging difficulties with verbal fluency tasks, the findings supported Hypothesis 1.

Tables 3 and 4 about here

3.2 Aim 2: Is verbal fluency performance related to executive functioning and/or language ability?

Hypothesis 2 (EF should be a stronger predictor of phonemic than semantic fluency; language ability should be a stronger predictor of semantic than phonemic fluency) and Hypothesis 3 (EF should show stronger relationships with performance in the later stages of each response trial; language ability should remain a predictor of performance throughout the response trial) were assessed by examining whether language and EF (ELWM, inhibition, switching) were related to verbal fluency performance. These analyses did not include group as a variable because this research question was more general and did not address group differences. Table 5 shows simple correlations between verbal fluency measures, age, non-verbal IQ, language ability, ELWM, inhibition and switching. Language ability and ELWM were significantly related to nearly all measures of verbal fluency (*r*s -.25 to .66), but there were few significant relationships between verbal fluency and inhibition (mainly with error scores, *r*s .23 to .33). There were no significant relationships between any aspect of verbal fluency and switching. Age showed a significant relationship with most semantic fluency measures (*r*s .32 to .50) and some

phonemic fluency measures (rs .26 to .34). Non-verbal IQ was unrelated to semantic fluency, but showed moderate relationships to many phonemic fluency variables (rs -.26 to .34).

Table 5 about here

Hypotheses 2 and 3 made specific predictions about which variables (language ability, ELWM, inhibition, switching) would account for significant amounts of variance in the verbal fluency measures. These hypotheses were assessed with further regression analyses in which age and IQ (control variables) were entered simultaneously with the four predictor variables. Tables 6 and 7 summarise key information from each regression predicting the following fluency measures: *response output* (total, valid), *output over time* (performance in quartiles 1, 2, 3, 4), *errors* (total errors, proportion errors), *switching*, and *cluster sizes*. Checks indicated that Mahalanobis distances were over 15 for 5 participants (1 SLI, 4 TD), therefore, these cases were omitted from regressions to avoid them having undue influence (Field, 2013).

<u>Aim 2 findings</u>: Language ability was a significant predictor of nearly all aspects of phonemic fluency performance (total and valid response output, performance in each quartile, switching, cluster size, words per cluster), and some aspects of semantic fluency (total and valid response output, performance in quartile 1). Inhibition was a significant predictor of error scores (total, proportion) in the phonemic fluency task, but these relationships were reduced to trends for the semantic fluency task (*ps*<.05). ELWM and switching were not significant predictors of verbal fluency performance (although a trend towards ELWM predicting numbers of switches in both tasks was apparent).

The overall percentage variance accounted for by each significant regression model ranged from 18-46%. No model accounted for more than half of the variance, implying that unmeasured variables and measurement error were also relevant.

Tables 6 and 7 about here

4. Discussion

4.1 Aim 1: What does verbal fluency performance reveal about children with language difficulties?

Verbal fluency tasks allow the investigation of children's' strategic search and generate processes, error monitoring, lexical/semantic/phonemic networks (Sauzéon et al., 2004), and controlled versus automatic processing. Although children with SLI are a heterogeneous group (Van Weerdenburg et al., 2006), and the current sample is likely to have had a range of receptive and/or expressive language difficulties, they nevertheless showed widespread difficulties on verbal fluency measures. Children with SLI were less able than typically developing peers to search semantic/phonemic networks and retrieve/generate items according to a rule. They showed weaknesses in automatic *and* controlled processing (early and later stages of response output), suggesting performance limitations for easily accessible items and items requiring effortful search strategies (Hurks et al., 2006). Children with SLI also made higher proportions of errors, suggesting they were less able to monitor their responses for accuracy and rule violations, and suppress incorrect responses. Further, they made fewer switches between sub-categories, which could imply less extensive or well-organised semantic/phonemic networks. These extensive verbal fluency difficulties in children with SLI supported Hypothesis 1.

Given the lack of robust evidence concerning effective interventions for children with language difficulties (e.g., Cirrin & Gillam, 2008; Law, Garrett, & Nye, 2004), the educational implications of these findings for interventions with children with SLI are speculative. Nevertheless, verbal fluency tasks tap highly relevant and generalizable classroom skills/abilities, as everyday learning activities require search, access, and retrieval of organised stored information according to relevant dimensions. Self-generation of comprehensive and accurate information is essential for many learning assessments: producing 'evidence' to support arguments in history or literature; answering fact-based scientific questions in biology, chemistry, or physics; using specific formulae/rules for carrying out mathematics operations; knowing spelling/grammar rules for language learning. The widespread difficulties of children with SLI compared to typical children with searching for and generating items from semantic/phonemic memory networks, and monitoring output for errors, could compromise everyday learning activities because access to stored knowledge is inefficient and error-prone. Interestingly, interventions that improve vocabulary and semantic processing/elaboration appear to have some effectiveness for school-age children (Cirrin & Gillam, 2008). However, because children with SLI showed weaker performance at all phases of the fluency tasks, including the more automatically generated first 15 seconds of the task during which it is assumed easily accessible items are generated (Hurks et al., 2006), it might be necessary to expand the range, depth and accuracy of semantic/phonemic knowledge/networks and consolidate new learning thoroughly so that it is 'automatically' available.

Difficulties with search, access and retrieval of information could be due to poor organisation, limited networks, or inaccurate semantic and phonemic knowledge, making search processes inefficient and error-prone. Children in the SLI group showed fewer switches between clusters of related items than typical children, implying reduced ability to perform exhaustive searches within a subcategory, and/or difficulties with the self-generation of new subcategories relevant to super-ordinate search categories. These findings could support the notion of reduced or poorly organised semantic/phonemic

networks (Nash & Snowling, 2008; Troyer et al., 1997). An alternative explanation is that children with SLI have adequate semantic/phonemic networks, but difficulties in accessing and generating relevant information quickly and efficiently due to processing speed limitations (e.g., Leonard et al., 2007). Cluster sizes on the phonemic fluency task were marginally smaller for children with SLI (*p*<.006), so even after identifying a subcategory of phonemically related items, search processes may be less productive. The relevant phonemic networks could have been reduced in size, phonemic information may have been inaccurate (there is evidence that phonological representations can be impaired for children with SLI, Marshall, 2014), or the neural associations between items within a category could have been weaker - our results cannot distinguish between these possibilities. However, this was a marginal result, and the sizes of stored subcategories relating to animal names did not differ between children with and without language impairments, so not all aspects of organised semantic knowledge were problematic (see also Marshall, 2014).

Children with SLI could be supported by imposing a clear structure on new information when learning, and promoting strong organisation and extensive/elaborate semantic/phonemic networks. Improving semantic/phonemic network organisation and accessibility may produce educational benefits in terms of more efficient storage, access and retrieval of information, with less scope for error. Elaboration strategies based on phonemic or semantic information have some support in the literature (Cirrin & Gillam, 2008). More speculatively, visual methods such as mind maps or colourful posters may offer non-verbal alternatives for the organisation of complex and detailed information, with key 'entry points' to facilitate access via easy to remember cues. Verbal fluency tasks are timed, and one limitation of the current study is that is not possible to know whether providing extra time could have enhanced performance, given that children with SLI have processing speed limitations (Leonard et al., 2007).

4.2 Aim 2: Is verbal fluency performance related to executive functioning and/or language ability?

There was limited evidence for the first part of Hypothesis 2, derived from Luo et al. (2010), that EF should be a stronger predictor of phonemic than semantic fluency (because phonemic fluency draws more heavily on EF skills). Inhibition scores related to errors on the phonemic fluency task (proportion and total errors), most likely because inhibition was required to monitor (and suppress) output for repeated/irrelevant items and rule violations. As there were no further significant relationships between any other EF variables and any other aspect of verbal fluency, this finding suggests that EF (i.e. inhibition) is a stronger predictor of phonemic than semantic fluency, but only in one area, namely error monitoring. However, although these data provide some support the first part of Hypothesis 2, a trend towards a relationship between inhibition and error scores on the semantic fluency task cautions against strong conclusions, and suggests that there may be a general inhibitory mechanism operating on error monitoring during both verbal fluency tasks.

The absence of relationships between ELWM and verbal fluency was unexpected, particularly given the simple correlations observed (see Table 5). It may be that relationships between ELWM and fluency disappeared because language ability was the stronger predictor in the regressions. Language ability and ELWM shared considerable variance (simple *r*=.69), and relationships between language ability and summary measures of executive functioning that include ELWM have been reported previously (Fitzpatrick, McKinnon, Blair, & Willoughby, 2014). Further, the lack of relationships between EF switching and verbal fluency might be surprising, given the role for switching within fluency tasks. However, switching between well-known sequences provided by the experimenter (e.g., letters, numbers), as in standard EF switching tasks (Trail Making), may not be the same as switching between self-generated clusters of items in verbal fluency tasks. These data do not support the suggestion (Diamond, 2013) that measures of verbal fluency are closely related to measures of EF task-switching.

The second part of Hypothesis 2, that language ability should be more related to semantic than phonemic fluency, was not supported. Language ability was based on expressive vocabulary, expressive language and semantic knowledge, and proved highly relevant for virtually all aspects of phonemic fluency performance, ranging from response output (total, valid, quartiles 1 through 4), to switching and cluster size. Only phonemic fluency errors (total, proportion) were unrelated to language ability. Thus, stronger language abilities played a role in the more difficult phonemic fluency measures throughout all quartiles of the task, and had an additional supportive role for performance on measures of cluster size and number of switches between generated sub-categories. For semantic fluency, verbal ability was related to total and valid measures of response output, and to performance in the first quartile of the task only. This suggests that better language ability supported performance in earlier parts of the semantic fluency task, when items were more readily available, but ceased to be important during the more effortful searching required during later parts of the task. Hence, no support was found for the second part of Hypothesis 2, that language ability should be a stronger predictor of semantic than phonemic fluency.

The first part of Hypothesis 3, that language ability should be related to fluency performance at all stages of the response trial (every quartile), was fully supported for phonemic fluency, but only partially supported for semantic fluency. This implies that Luo et al. (2010) were correct to surmise that language ability is a fundamental skill underlying phonemic fluency performance, regardless of whether we consider items generated in the early 'burst' at the beginning of the task, or later items when performance levels drop off and search processes become more demanding (Hurks et al., 2006). However, effortful searching for items towards the middle and end of the semantic fluency task did not appear to be supported by language ability. The second part of Hypothesis 3, that EF should show stronger relationships with performance towards the end of each response trial, was not supported for either fluency task. No relationships between EF variables and performance in the four quartiles of the

task were found. It was not possible, therefore, to draw conclusions about whether EF skills are recruited more heavily for effortful later portions of the task as opposed to earlier automatic searching. The current findings would suggest that such relationships are not apparent.

One final issue concerns whether these results for a mixed sample of children with a wide range of language and EF abilities would generalise to typical samples. Exploratory regressions on the typical sample alone indicated this was the case. Most findings were very similar in the typical group, with relationships between fluency and language ability, and links between inhibition and error scores.

5. <u>Conclusion</u>

Children with SLI showed widespread difficulties with multiple aspects of verbal fluency performance relative to typical children, including searching, retrieving and generating words according to a rule, switching between relevant semantic and phonemic sub-categories, and monitoring output for errors such as repeated items and rule violations. These difficulties were apparent for easily accessed items at the beginning of the task ('automatic processing'), and later items requiring more demanding search efforts.

Two factors identified in previous literature as predictors of verbal fluency performance were explored: language ability and EF (ELWM, inhibition, switching). Language ability related to virtually all aspects of phonemic fluency performance and some aspects of semantic fluency performance. The only measure of EF to show significant relationships with verbal fluency was inhibition, which related to error monitoring in the phonemic fluency task. Overall, the relationships between verbal fluency and EF were modest compared to the relationships between fluency and language ability. The findings suggest that verbal fluency difficulties in children with language impairments reflect language weaknesses, with a contribution from executive inhibition difficulties in relation to error monitoring.

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Table 1: Mean scores (SD in brackets) and ranges (lower line) for descriptive data for all groups.

Variable/Group	SLI (n=41, 28 boys)	Typical (n=88, 59 boys)		
Age (years; months)	11;6 (16m)	9;10 (28m)		
	(8;1–14;1)	(6;0-14;8)		
BAS-II Matrices T-score ¹	54.6 (6.2)	57.4 (6.9)		
(Non-verbal IQ, NVIQ)	(46-71)	(40-78)		
BAS-II Verbal ability ²	100.8 (18.3)	111.5 (21.5)		
	(54.5-136.5)	(65-160)		
Age equivalent average	7;9 (13m)	10;3 (33m)		
score⁴ (years; months)	(6;0-10;3)	(6;7-15;11)		
Recalling sentences ³	5.2 (2.5)	10.4 (1.9)		
(CELF-4-UK)	(1-10)	(8-15)		
Formulated sentences ³	3.8 (2.5)	10.5 (1.9)		
(CELF-4-UK)	(1-8)	(8-14)		
Word classes receptive ³	5.2 (1.6)	10.2 (2.0)		
(CELF-4-UK)	(1-7)	(8-15)		
Word classes expressive ³	5.7 (1.8)	10.7 (1.8)		
(CELF-4-UK)	(1-9)	(8-15)		

¹ T-scores mean=50, SD=10

² Average ability scores - equivalent to raw scores – mean across Word Definitions and Verbal Similarities

³Scaled scores mean=10, SD=3

⁴Average age equivalent score across the four CELF subscales (note these are estimates)

Table 2: Mean Scores (SD) and ranges for verbal fluency variables.

Variable/Group	SLI (n=41, 28 boys)	Typical (n=88, 59 boys)
Semantic fluency total	31.05 (6.88)	34.37 (7.47)
,	21-47	17-55
Semantic fluency valid	30.17 (6.62)	33.94 (7.53)
	21-46	17-55
Quartile 1	11.68 (2.53)	13.24 (3.15)
	6-19	5-19
Quartile 2	7.46 (2.13)	8.23 (2.24)
	3-13	3-14
Quartile 3	5.80 (2.37)	6.57 (2.50)
	1-11	2-15
Quartile 4	5.20 (2.40)	5.76 (2.60)
	1-12	0-14
Total errors	.88 (.95)	.43 (.71)
	0-3	0-3
Proportion errors	.03 (.03)	.01 (.02)
	.0012	.0012
Number switches	7.61 (3.20)	8.97 (3.08)
	1-17	4-21
Words per cluster	2.01 (.76)	1.87 (.44)
	1.17-5.50	1.17-3.20
Phonemic fluency total	23.12 (8.83)	33.53 (9.89)
	(8-49)	(13-60)
Phonemic fluency valid	20.46 (7.72)	31.65 (10.11)
,	(7-45)	(12-59)
Quartile 1	9.44 (2.99)	12.20 (3.75)
	(5-19)	(4-21)
Quartile 2	4.34 (2.60)	7.78 (3.21)
	(1-11)	(2-16)
Quartile 3	3.90 (1.95)	5.88 (2.69)
	(0-9)	(1-13)
Quartile 4	2.98 (2.17)	5.81 (3.05)
	(0-9)	(0-14)
Total errors	2.66 (3.28)	1.88 (1.81)
	(0-17)	(0-8)
Proportion errors	.11 (.11)	.06 (.06)
	(.0053)	(.0028)
Number switches	14.68 (6.49)	20.80 (6.99)
	(4-37)	(8-46)
Words per cluster	1.31 (0.18)	1.42 (0.23)
	(1.08-1.74)	(1.00-2.05)

Table 3: Regression summaries: **semantic fluency**. Two predictor variables were entered at Step 1 (age, non-verbal IQ). The dummy-coded group variable was entered at Step 2 (SLI-vs-typical). Information about *Step 2* includes total variance accounted for (total R^2), standardised beta-values for each predictor variable, and change in R^2 . Significance values indicated where relevant. Group differences highlighted in bold. N=129.

Semantic Fluency	Total R ²	Details of Step 2 for each regression (when 'group' dummy variables						
Measure	accounted for	entered)						
	by the model							
		ΔR^2 Step 2	β Age	βΝνία	β SLI-v-			
					Typical			
Total responses	.43***	.16***	.66***	.08	.43***			
Valid responses	.47***	.18***	.67***	.09	.47***			
Quartile 1	.35***	.14***	.56***	.13	.42***			
Quartile 2	.22***	.09***	.47***	.04	.32***			
Quartile 3	.30***	.16***	.36***	.23**	.44***			
Quartile 4	.33***	.24***	.38***	.12	.54***			
Total errors	.09**	.06**	13	13	27**			
Proportion errors	.11**	.07**	17	13	29**			
Number switches	.22***	.11***	.45***	.05	.36***			
Words per cluster	.01	.01	08	.04	09			

*p<.05; **p<.005; ***p<.001. Alpha was set at p<.005 after Bonferroni corrections

Table 4: Regression summaries: **phonemic fluency**. Two predictor variables were entered at Step 1 (age, non-verbal IQ). The dummy-coded group variable was entered at Step 2 (SLI-vs-typical). Information about *Step 2* includes total variance accounted for (total R^2), standardised beta-values for each predictor variable, and change in R^2 . Significance values indicated where relevant. Group differences highlighted in bold. N=129.

Phonemic Fluency	Total R ²	Details of Step 2 for each regression (when 'group' dummy variable						
Measure	accounted for	entered)						
	by the model							
		ΔR^2 Step 2	β Age	β Ν νια	β SLI-v-			
					Typical			
Total responses	.44***	.29***	.48***	.17*	.60***			
Valid responses	.51***	.33***	.50***	.22**	.63***			
Quartile 1	.42***	.20***	.52***	.22**	.49***			
Quartile 2	.36***	.28***	.36***	.13	.58***			
Quartile 3	.30***	.16***	.36***	.23**	.44***			
Quartile 4	.33***	.24***	.38***	.12	.54***			
Total errors	.09*	.02	22*	17	17			
Proportion errors	.18***	.08**	30**	20*	30**			
Number switches	.37***	.23***	.46***	.17*	.52***			
Words per cluster	.07*	.06**	.06	.02	.27* ^a			

p*<.05; *p*<.005; ****p*<.001; ^a*p*=.006. Alpha was set at *p*<.005 after Bonferroni corrections

Table 5: Correlations between fluency, age, non-verbal IQ, language ability and executive functioning measures (ELWM, inhibition, switching). N=129.

Variables	Age	NVIQ	Language ability	ELWM	Inhibition	Switching
Semantic Fluency						
Total responses	.50**	.16	.64**	.56**	21	.03
Valid responses	.50**	.18	.66**	.58**	24*	.04
Quartile 1	.40**	.21	.56**	.45**	20	.16
Quartile 2	.35**	.10	.48**	.36**	17	08
Quartile 3	.37**	.19	.47**	.49**	15	03
Quartile 4	.33**	.01	.37**	.37**	14	.02
Total errors	03	17	15	20	.23*	09
Proportion errors	07	19	20	25*	.26*	09
Number switches	.32**	.12	.46**	.45**	12	.00
Words per cluster	.04	03	06	09	.00	.10
Phonemic Fluency						
Total responses	.26*	.29*	.59**	.55**	12	.09
Valid responses	.27*	.34**	.63**	.57**	18	.09
Quartile 1	.34**	.32**	.64**	.56**	22	.02
Quartile 2	.15	.24*	.47**	.41**	05	.09
Quartile 3	.20	.31**	.47**	.40**	10	.15
Quartile 4	.18	.22	.45**	.49**	20	.06
Total errors	16	20	26*	19	.29*	06
Proportion errors	19	26*	39**	30**	.33**	05
Number switches	.27	.27*	.52**	.53**	12	.02
Words per cluster	03	.07	.20	.05	02	.13

Table 6: Regression summaries predicting **semantic fluency**. Six predictor variables were entered in a block (age, non-verbal IQ, language ability, ELWM, inhibition, switching). Information provided includes total variance accounted for by the model (total R^2) and standardised beta-values for each predictor variable. Significance values indicated where relevant. N=124.

Semantic Fluency	Total R ² accounted for	Standardised	Beta-values				
Measure	by the model	Age	NVIQ	Language ability	ELWM	Inhibition	Switching
Total responses	.43***	.12	05	.40**	.18	08	04
Valid responses	.46***	.10	04	.41***	.20*	11	04
Quartile 1	.33***	.08	.00	.43**	.07	09	.17*
Quartile 2	.25***	.06	01	.35*	.10	04	09
Quartile 3	.24***	.14	.05	.16	.18	05	16
Quartile 4	.18**	.06	12	.15	.23	07	07
Total errors	.10	.13	06	09	12	.21*	07
Proportion errors	.13*	.13	06	11	17	.22*	08
Number switches	.22***	.03	.00	.22	.25*	01	06
Words per cluster	.03	.12	03	04	09	01	.10

p*<.05; *p*<.005; ****p*<.001. Alpha was set at *p*<.005 after Bonferroni corrections

Table 7: Regression summaries predicting **phonemic fluency**. Six predictor variables were entered in a block (age, non-verbal IQ, language ability, ELWM, inhibition, switching). Information provided includes total variance accounted for by the model (total R^2) and standardised beta-values for each predictor variable. Significance values indicated where relevant. N=124.

Phonemic Fluency	Total R ² accounted	Standardised	beta-values				
Measure	for by the model	Age	NVIQ	Language ability	ELWM	Inhibition	Switching
Total responses	.35***	25*	.02	.60***	.15	01	02
Valid responses	.44***	28**	.06	.66***	.13	08	02
Quartile 1	.42***	12	.05	.59***	.08	12	07
Quartile 2	.26***	31*	.02	.58***	.12	.05	.05
Quartile 3	.22***	19	.13	.52***	01	02	.01
Quartile 4	.25***	27*	02	.41**	.21	14	06
Total errors	.15* (<i>p</i> =.005)	.03	12	22	.08	.26**	04
Proportion errors	.23***	.10	12	36*	.01	.26**	03
Number switches	.29***	13	.02	.40**	.24*	01	06
Words per cluster	.11*	31*	.00	.51**	23	02	.08

p*<.05; *p*<.005; ****p*<.001. Alpha was set at *p*<.005 after Bonferroni corrections