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Category specific recall in acute stroke: a case with letter speech

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ABSTRACT

Category selective recall in spontaneous speech after stroke has been reported only rarely. We recently described three cases demonstrating transient number speech in the acute stage of left hemispheric stroke and hypothesized a link with multilingualism and mathematical proficiency. In this report, we describe a similar case with a transient episode of utterances of randomly selected letters. Like in the three previous cases, this episode was preceded by a brief stage of mutism and ultimately evolved to Wernicke's aphasia over a period of days. This phenomenon is reviewed with reference to linguistic models and neuroanatomic and neurophysiological correlates.

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Introduction

The existence in aphasic patients of category-specific word-finding problems, i.e. an anomia for one domain of semantic knowledge relative to others, has challenged our concepts of semantic organization. As can be expected from the continuing debate on the localizational versus holistic nature of language organization, two opposing theories have emerged as an explanation for category-specific deficits: the domain-specific and domain-general thesis (Reilly, Rodriguez, Peelle, & Grossman, 2011). The domain-specific theory suggests a true anatomically demarcated localization of specific semantic categories (Caramazza & Mahon, 2003, 2006; Hillis, Rapp, & Caramazza, 1995; Laiacona, Capitani, & Caramazza, 2003; Mahon & Caramazza, 2009), while the domain-general theory argues for an organization of categories as a function of their sensory, motor or functional properties (Farah & McClelland, 1991; Gonnerman, Andersen, Devlin, Kempler, & Seidenberg, 1997). Both theories are consistent with the inability to retrieve words from one category as a consequence of damage to a specific cerebral localization. For example, the inability to recall the domain-general "operative items" which are learned through both visual and sensorimotor experience are related to an occipito-parietal area dysfunction. An impairment for the recognition of a domain-specific category, e.g. "animals", is related to damage to the inferior temporal cortex (Ferreira, Giusiano, & Poncet, 1997).

In contrast to deficient word finding for one semantic category, the opposite phenomenon in which only one category is preserved while all other verbal semantic categories are suppressed has been described only rarely. De Letter et al. (2012) described three patients who demonstrated number speech in the hyperacute stages of aphasia after stroke. During a short period of time after stroke onset, these patients uttered randomly selected numbers as the only category of items. Number speech manifested in three multilingual and mathematically educated men as a transitional stage between initial mutism and the eventual development of Wernicke's aphasia. The development of this category-specific production was assigned to the combination of a large lesion in the left temporo-parietal cortex and the background in mathematics of the patients. It is not evident to position this finding within the frameworks of semantic organization, since number processing and meaning are tightly linked to aspects of magnitude, scale, serial order and spatial representation (Fias & van Dijck, 2016). However, numbers can also be considered lexical elements within a specific semantic class. We hypothesized that, in the three patients with number speech, semantic organization had developed with a strong link between domain-general properties and lexical access to numbers as a consequence of a training in mathematics, leading to a favoring of utterances from this specific class of elements when recovering from an initial stage of mutism and in the presence of severe Wernicke's aphasia.

The current report describes a similar manifestation in a patient displaying letter speech after stroke, characterized by the preferential utterance of randomly selected letters. To our knowledge, isolated letter speech as a transitory phenomenon preceding Wernicke's aphasia has never been described before. Letter and number speech can be considered two comparable phenomena in which elements of specific semantic categories, sharing the characteristic of automatic series, are uttered preferentially in the hyperacute stages of stroke and following a brief stage of mutism. The terms "automatic" and "non-propositional" speech were first introduced by Baillarger (1865) and Hughlings Jackson (1879) to denote automatically and involuntarily produced overlearned utterances. Ordinal, non-propositional (automatic) categories are, for example, counting, reciting the days of the week or the months of the year and reciting the alphabet. Their processing shows an

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activation bias toward the right fronto-temporo-parietal network (Eagleman, 2009). The non-ordinal categories (such as names of furniture, animals, cars, and fruit) are part of propositional speech and are preferentially processed in the left hemisphere (Eagleman, 2009). The production of letter and number series can, therefore, be classified in the category of ordinal, non-propositional (automatic) speech. However, in our patients numbers and letters were produced randomly.

In this report letter speech and its evolution will be described and compared with number speech in the context of existing models of semantic organization and neurophysiological alterations in the early stages of stroke.

Methods

Patient

HR is a 63-year-old right-handed (Van Strien, 1992) technical engineer who was admitted to the hospital after a wake-up ischemic stroke. Since the exact time of stroke onset could not be determined, HR was not eligible for thrombolytic therapy. His medical history mentioned a myocardial infarction in 1998, an episode of ventricular fibrillation in 2013 and a minor uncomplicated traffic accident in 2014. His native language was Dutch with a high proficiency in German, French and English as second, third and fourth languages, respectively. HR practiced his second language every week during professional and leisure activities.

At the neurological examination in the emergency ward HR presented with mutism, severe hemiparesis of the right arm and leg (arm and leg both 1/5 on the MRC grading scale), right facial paresis and hypoesthesia and a gaze deviation to the left side. CT imaging confirmed the presence of a large ischemic lesion in the left hemisphere, encompassing the insula and the temporo-parietal cortex, reaching from the centrum semiovale to the corona radiata . Due to an extracranial metallic particle as a consequence of his traffic accident some years before, MR-scan was impossible (Figure 1). Unfortunately, functional neuroimaging with SPECT of PET scan was not performed.

After 2 to 3 h of initial mutism, HR demonstrated letter speech (randomly selected letters from the Dutch alphabet) along with severe comprehension difficulties. One day post-onset, jargon aphasia set in, interrupted by letter speech and "polyglotte reaktionen" such as "We don't *hoef* them all" (*hoef*= Dutch for "need") or "Ik kan de bestelling *wechseln*" ("I can change the order"; *wechseln* is a German word instead of a Dutch one). Subsequently, HR developed a severe anomia accompanied by perseverative answers. At the end of the first week, letter speech had disappeared and jargon aphasia had installed.

Extensive investigation allowed to identify atrial fibrillation with cardiac embolism as the underlying cause of his stroke. Echographic evaluation of the carotid and vertebral arteries suggested some atherosclerotic plaque formation without hemodynamic impairment in both carotid arteries. The patient was started on a treatment with oral anticoagulation.

Procedure

Neurolinguistic evaluation

Due to the scheduling of necessary medical investigations, the unexpected presentation of this syndrome and therefore the unprepared circumstances in which the informed consent and the speech investigations had to be performed, complete, standardized and extensive data sampling were impossible. As we could not estimate how long the letter speech would persist, analysis was performed post-hoc based on clinical video-recordings of speech and language.

Week 1. In the first hours after stroke, with the alternation of mutism and letter speech, the patient was investigated by means of the ScreeLing (Visch-Brink, van de Sandt-Koenderman & El Hachioui, 2010), a screening instrument that examines semantic, phonologic and syntactic processing. Since auditory comprehension was severely disturbed, standardized screening was very difficult. Video-recorded language samples (spontaneous speech, repetition, naming, comprehension, automatic series, reading and writing) were collected to describe and analyze the letter speech phenomenon in the first hours and days post onset. In every modality, the amount of words spoken by the patient was counted. When the "letters" were produced in the correct order of an existing word, the "letters" were interpreted as a word. The number of letter productions was divided by the total number of real words, non-real words and letters, resulting in a percentage letter speech. In order to observe the evolution of letter speech, the modalities "spontaneous speech" and "writing" were video-recorded and transcribed at 1, 5 and 8 days post-stroke.

Week 2. Two weeks after stroke onset the patient's aphasia was stabilized and HR was able to undergo a standardized language examination. At that time, all subtests of the complete Dutch version of the standardized Aachen Aphasia Test (AAT) (Graetz, De Bleser, & Willmes, 1992) were administered as

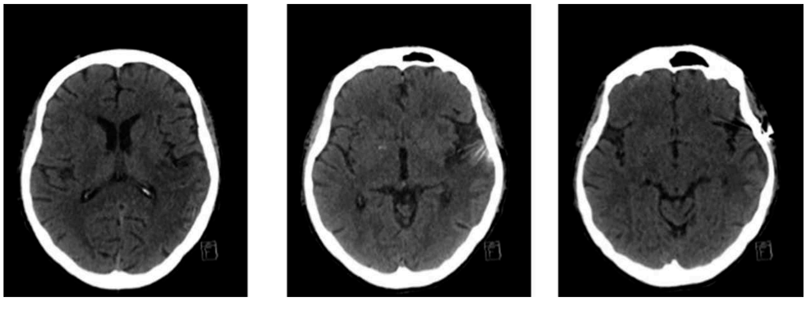

Figure 1. CT scan of the brain, demonstrating the presence of an extranial metallic particle and a large hypodensity in the left hemisphere.

well as three auditory and two visual subtests of the Dutch version of the Psycholinguistic Assessment of Language Processing in Aphasia (PALPA) (Bastiaanse, Bosje, & Visch-Brink, 1995). The PALPA phoneme discrimination tests consist of a same-different judgment task in which the patient has to judge whether pairs of pseudowords (PALPA 1) or minimal pairs of real words (PALPA 2) are the same or not. The patient was instructed to say "yes" as a response to a similar word pair and to say "no" to a dissimilar word pair. The auditory stimuli are monosyllabic (pseudo)words of consonant/vowel/consonant (CVC)-structure. They all differ in phonemic contrasts, voicing, place of articulation (PoA) or manner of articulation (MoA), which are either initial, final or based on metathesis (i.e. an altered sequence of phonemes). The auditory lexical decision task in subtest PALPA 5 contains 80 real words and 80 pseudowords. The pseudowords are derived from the real words by changing one or multiple phonemes, making them genuine pseudowords. For each stimulus HR was asked to decide whether it was a real word or not, by saying "yes" to a real word or "no" to a pseudoword. The visual lexical decision task (PALPA 24) has the same structure and words as the auditory version, but now the words are presented visually and HR had to cross out all pseudowords. The matching of capital letters with small letters (PALPA 18) requires the combination of 26 capital letters to the corresponding small letter. The patient is asked to choose between two small typed letters.

Speech intervention started as soon as the patient was admitted to the hospital. After a short screening of the different language modalities, speech rehabilitation focussed on eliciting spontaneous speech, in combination with comprehension tasks. The speech-language pathologist used oral and written language simultaneously in all language modalities.

Statistical analysis

Descriptive statistics was performed in order to unravel the letter speech phenotype and the relationship with jargon aphasia.

Results

Neurolinguistic evaluation

Day 1 (day of the wake-up stroke): letter speech

(1) Letter Speech

During the first day after stroke, HR presented with mutism and severe difficulties in understanding simple yes/no questions. His pragmatic communication behavior was relatively spared, including eye contact, gesticulations, turn taking and head nodding. Some hours after stroke onset letter speech appeared and was soon alternated with the production of real words. Letters were randomly selected and were unrelated to a target word. They were pronounced as in the Dutch alphabet: [a:] for A, [se:] for C, [e:] for E, [te:] for T, [ɛl] for L and [ɛs] for S.

"Patient: I have not experienced yet, a difficult word. I thought maybe a possible opson euh A, C, E, C, E, T, E, E, E, L, E, L, S, L as a as Ooo, swol, smel, A, C, B, C, E, then? And then I can probably A, C, E, C, E, C, E, euh, E, L, E, E, C, E, L."

During the first day, the patient remained unable to repeat, to write or copy his own name or to read regular words in a comprehensive way. Letter speech was present in spontaneous speech (51%), reading (82%), repetition (79%), auditory comprehension (66%), naming (40%) and automatic speech (60%). Error analysis demonstrated neologisms in all language modalities except for the naming task where perseverations on word and sentence level and slight information transfer could be observed. No "polyglotte reaktionen" were observed at this stage of recovery (Table 1).

(1) Letter speech, compared with number speech and jargon aphasia on day 1

The linguistic context (e.g. fluency, type of aphasia, recovery pattern, paraphasias, prosody, auditory feedback) in which letter speech occurred as well as the lesion localization and education of the patients were similar in number speech and jargon aphasia. Topics on which letter and number speech differed from jargon aphasia were the short, transitional duration of the phenomenon, the spontaneous speech with random selection of elements from automatic categories and perseverations (Tables 2 and 3).

Day 2: language production

A decrease of letter speech was observed in spontaneous speech (28%) on day 2. Like on day 1, letter speech occurred especially during reading (71%), repetition (68%) and auditory comprehension (88%). Automatic series could be produced perfectly and even in naming letter speech occurred only (19%) when the patient lost attention. Neologisms at word level were more present than on day 1. Spontaneous speech was characterized by semantic paraphasias and "polyglotte reaktionen" (Table 4).

When the patient was asked to write down his name, HR was able to use pen and paper, but the result was a destructured

Table 1. Letter speech in six language modalities, produced by HR on day 1.

	Total number of words	Number of real words	Percentage of letter speech (count)	Error analysis (count)
Spontaneous speech	37	19	51 (16/37)	Neologism (2)
Reading	28	1 (improvised)	82 (23/28)	Neologism (4)
Repetition	34	0	79 (27/34)	Neologism (7)
Auditory comprehension	12	0	66 (8/12)	Neologism (4)
Naming of body limbs, pointed by the therapist	50	30	40 (20/50)	Perseverations on word and sentence level + slight information transfer
Automatic speech	15	4	60 (9/15)	Neologisms (2)

Table 2. Comparison of the linguistic and environmental context of letter speech in the spontaneous speech modality with the linguistic context of number speech (De Letter et al., 2012) and jargon aphasia (literature).

	Letter speech (*n* = 1)	Number speech (*n* = 3)	Jargon aphasia
Speech characteristics			
Short, transitional duration (hours to days) of the phenomenon	x	x	0
Non-propositional speech	x	x	0
Propositional speech	slightly	slightly	x
Spontaneous speech with random selection out of automatic series	x	x	0
Fluent, well-articulated but unintelligible speech	x	x	x
Related to Wernicke's aphasia (pressure of speech, empty speech, logorrhea, intact prosody)	x	x	x (severe)
Phonemic and semantic paraphasias or neologisms	x	x	x
No auditory feedback	x	x	x
Perseverations	x	x	-
Polyglotte Reaktion	x	x	-
Euphoria	-	x	-
Non-speech characteristics			
Profession	Technical engineer	Civil engineers	Not specified
Stroke localization	Temporo-parietal	Temporo-parietal	Temporo-parietal

Table 3. Speech in several language modalities, produced by HR on day 2.

	Total number of words	Number of real words	Percentage of letter speech (count)	Error analysis (count)
Spontaneous speech	318	224	28 (88/318)	Semantic paraphasia (1) Neologisms (6) "Polyglotte reaktion" in L1 (1)
Reading	52	3	71 (37/52)	Neologism "C,E,L" (12)
Repetition	62	1	68 (42/62)	Neologism "C,E,L" (19)
Auditory comprehension	17	0	88 (15/17)	Neologism "C,E,L" (2)
Naming of colors, shown by the therapist	32	25	19 (6/32)	Neologism (1)
Non-propositional (automatic) speech	9	8	0 (0/9)	Neologism (1)

Table 4. Spontaneous speech on day 5 and day 8 post-stroke.

	Total number of words	Number of real words	Percentage of letter speech (count)	Error analysis (count)
Day 5	183	170	0	Phonemic paraphasia (6) "Conduite d'approche" (5) Neologism (13) Echoing approval (2)
Day 8	40	30	0	"Conduite d'approche" (5) "Conduite d'ecart" (4) Neologism (10) Phonemic paraphasia (5) "Polyglotte Reaktion" in L2 (1)

reproduction of real and non-real letters and stripes (Figure 2). Writing looked as if the patient was trying to "draw" graphemes. He was unable to synthesize graphemes into words.

Day 5: language production

In order to minimize the interference with the speech rehabilitation, the linguistic evaluations on day 5 and 8 were limited to a video-recorded spontaneous speech sample and a writing task. On day 5, letter speech had completely disappeared and there was evidence of phonologic and lexico-semantic recovery. Proportionally, the number of neologisms (13) had increased, accompanied by phonemic paraphasias (6) and "conduite d' approches" (5). The global information transfer was poor. "Polyglotte reaktionen" had disappeared (Table 5).

The writing skills had improved substantially, probably driven by the written support during oral speech rehabilitation. Drawing was replaced by writing in capital letters which, however, were sometimes mirrored, as can be seen in Figure 2.

Day 8: language production

On day 8 the spontaneous speech contained more information. Word finding problems impaired spontaneous speech, captured by phonemic paraphasias (5), sometimes within a "conduite d' approche" (5) or "conduite d' écart"(4), neologisms (10) and one "polyglotte reaktion" in L2.

The writing abilities had further improved. The patient no longer wrote in mirror images. However, in written reproduction orthographic selection and sequencing problems persisted (Figure 2).

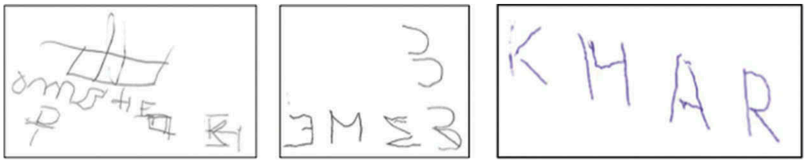

Figure 2. Patient trying to write his own name on day 2, day 5 and day 8, respectively, from left to right.

2 weeks (15 days) post-onset

Two weeks post-onset, the aphasia severity was stable, making it possible to examine the patient with standardized neurolinguistic tests.

The AAT (Graetz et al., 1992) concluded to a pure (100%) and severe Wernicke's aphasia with repetition (47th percentile), reading and writing (21st percentile), naming (47th percentile) and comprehension (29th percentile) problems (see Table 5). The token test (16th percentile), which is considered an equivalent for aphasia severity in the AAT, was difficult due to perseverations and a disturbance of divided attention between colors and forms. In the first repetition module, HR tried to form words with the letters presented, sometimes resulting in neologisms. Reading was impaired, due to a lack of attention for grapheme-phoneme conversion, resulting in guessing. Writing letters without mirroring was possible. However, letters were written randomly and could not be synthesized to a word, resulting in letter writing and perseverations of those letters (Table 5).

Since the phonological disabilities prevailed in all language modalities, the Dutch version of the PALPA (Bastiaanse et al., 1995) was conducted for 1) auditory discrimination for non-real words and 2) minimal pairs, 3) orthographic discrimination (matching capital letter with lowercase), 4) auditory lexical decision (imageability and frequency) and 5) orthographic lexical decision (imageability and frequency). Although all these tests were mildly impaired, a better recovery was observed for auditory (≥91%) than for orthographic input processing (≤77%). Within the orthographic input processing, lexical decision (69%) was more impaired than orthographic discrimination. Especially the non-real words were judged as real words by the patient. Within the slightly disturbed judgment of real words, the two words considered as non-real words did not carry specific characteristic of imageability or frequency, neither in auditory nor in orthographic lexical decision (Table 5).

Discussion

The current manuscript reports on a case of random letter speech production as the predominant speech production in the first hours after a large left hemispheric ischemic stroke, following a brief period of initial mutism. Spontaneous speech in the first hours after stroke was a concatenation of randomly selected letters, occasionally interrupted by a meaningful word or a phonological neologism.

Linguistic testing was compromised by the setting of a hyperacute stroke, with several medical interventions that interfered with more refined analysis. However, we found that randomized letter speech occurred in all language modalities (spontaneous speech, reading, writing, repetition, naming and rehearsal of automatic series), with the highest prevalence in reading, repetition and auditory comprehension (as a reply to instructions). The letter production phenomenon was accompanied by severe auditory and written comprehension deficits. Although the preverbal purpose of the message could not be understood, the verbal utterances seemed to be produced in a fluent way, with respect to the phonotactic (syllable structure, consonant clusters, vowel consequences) and prosodic (intonation, dynamic variation, accentuation, rhythm) rules of Dutch (native) language.

Letter speech evolved toward an undifferentiated jargon aphasia and subsequently neologistic jargon aphasia within the course of 1 week after stroke onset. In fact, the pressure driving letter speech can be compared to logorrhea in jargon aphasia, which was described by Alajouanine (1956) as a "pressure of speech" in which the patient "showed indisputably a lack of voluntary influence". Jargon aphasia is considered a difficulty with understanding language while the verbal expression is unintelligible due to minimal content (Alajouanine & de Ribaucourt, 1952; Marshall, 2006). Although

Table 5. Results on the Aachen Aphasia Test (Graetz et al., 1992) and Psycholinguistic Assessment of Language Processing in Aphasia (PALPA) (Bastiaanse et al., 1995).

Aachen Aphasia Test

Language modality	Score	Percentile (%)
Token Test (/50)	3	16
Repetition (/150)	104	47
Reading and writing (/90)	17	21
Naming (/120)	74	47
Auditory and visual comprehension (/120)	70	29

Psycholinguistic Assessment of Language Processing in Aphasia

Language modality	Score (%)	Median of norm group	Error analysis (count)
Auditory discrimination - nonreal words (/72)	70 (97)	70.05 (± 1.64)	- Methatesis (1) - Articulation place (1)
Auditory discrimination – minimal pairs (/72)	66 (91)	70.80 (± 1.94)	- Final phoneme (2) - Initial phoneme (1) - Metathesis (2) - Place of articulation (3) - Manner of articulation (1)
Orthographic discrimination - Matching Capital letter-lowercase (/26)	20 (77)	25.83 (± 1.09)	
Auditory lexical decision- imageability and frequency (/160)	154 (96)	158.25 (± 2.27)	- nonreal words (4) - real words (2): imageability (1high, 1 low) + frequency (1 high, 1 low)
Orthographic lexical decision-imageability and frequency (/160)	110 (69)	158.72 (±1.78)	- nonreal words (58) - real words (2): high imageability (2) + high frequent (1) and low frequent (1)

spontaneous production is fluent and well articulated with intact syntactic form and prosody, the speech is empty (Butterworth, 1985), logorrheic and characterized by phonemic and semantic paraphasias and neologisms (jargon) (Alajouanine, 1956). The characteristics of letter speech are therefore in accordance with the development of jargon aphasia.

The evolution during the initial stages, as well as the subsequent evolution during the first year after stroke were highly comparable to the evolution in our previously reported three patients with number speech in the acute phases of stroke (De Letter et al., 2012). Number speech also occurred as a transient phenomenon between mutism and fluent aphasia in the context of a large hemispheric stroke in peri-insular and temporoparietal areas. The context in which number speech occurred coincided with the linguistic findings in the case of letter speech reported here. Similar to our number speech patients, our letter speech case evolved progressively from a neologistic jargon aphasia toward a mild semantic jargon aphasia over the course of months, which is considered a normal evolution in undifferentiated aphasia (for a review see Buckingham, 1981). The eventual severity of aphasia in our number speech cases was variable in the long run.

Letters and numbers belong to the category of non-propositional (automatic) speech and are usually associated with neural activity in right hemispheric networks. Propositional (non-automatic) speech is rather related to left hemispheric activation. Robinson et al. (2015) distinguish between at least four mechanisms involved in the early conceptual preparation stage for producing left-lateralized propositional (non-automatic) speech. The first three mechanisms involve the generation of novel thoughts, the sequencing of novel thoughts, and the selection among competing thoughts. These first three mechanisms are typically disturbed in patients with dynamic aphasia. The fourth mechanism contrasts with the others by its inhibitory/excitatory regulation of the first three mechanisms. A disturbance of the fourth mechanism causes a quick and continuous flow of propositional speech and a failure in stopping the generation of novel thoughts (logorrhea), which is typically present in jargon aphasia. However, if letter (and number) speech would simply be a logorrhea-induced recall of non-propositional (automatic) speech, and as such a precursor of jargon aphasia, one would expect the patients to produce their letters and numbers in a serial order (A,B,C,D,E or 1,2,3,4). Contradictorily enough, all patients produced their letters and numbers in a randomized order. Random selection of items can be explained by the fact that only one category is activated, especially the category that requires the least retrieval effort. Therefore, perseverative utterance of randomly selected letters can be explained by a combination of the "spew" and "echo box" hypotheses:

- The "spew" hypothesis (Underwood & Schulz, 1960) assumes that high-frequency items tend to be produced first in free responding situations. This hypothesis posits that items are not retrieved as independent but rather as interdependent units, so that the recall of one item affects the recall of the next. This recall for units can exceed the span, which is explained in terms of the unitization of units into higher-order structures, which in turn are called higher-order units (Paivio & Walsh, 1993). For the English language, high frequent consonants are C, D, F, H, L, R, S and T, while low frequent consonants are B, G, J, K, P, Q, V, X and Z (Underwood & Schulz, 1960). The letters selected by our patient are all high frequent (higher-order) letters in Dutch, which could explain why they are randomly recalled.

The "echo box" hypothesis is based on the empirical finding of a recency effect in free recall (Murdock, 1962), combined with a rapid decay of the acoustic trace of recently presented items in the working memory (Sperling & Speelman, 1970), later defined as the "articulatory loop" (Baddeley & Della Sala, 1996). This recency effect could explain the perseverations in the patients' letter recall.

When considering these hypotheses on item retrieval, it is necessary to take into account the issue of plasticity and formation of memory traces during development and education. The current patient with letter speech, as well as the three patients previously described with number speech (De Letter et al., 2012), all have a mathematical background (engineering) and are all educated multilingually. The fact that they share a mathematical formation may intuitively be regarded as important in the context of number speech, since numbers may be considered as highly frequent items in their vocabulary. This does not apply to letter speech, however. Mathematical giftedness has been studied extensively, but the results of various studies investigating cerebral processing of different tasks have been conflicting and task-dependent. A study of crossed aphasia patients with acalculia allowed concluding that language and calculation share the same hemispheric dominance, which might suggest common underlying computational systems (Semenza et al., 2006). Multilinguals develop more control abilities because their multilingualism forces to control the continuous interference between lexical representations of the two languages, which has even been suggested to reduce the odds of developing neurodegenerative disorders (Bialystok, Craik, & Luk, 2008; Woumans et al., 2015). Speech rehabilitation after stroke could entail generalization from one language to another (untrained) language (Edmonds & Kiran, 2006; Filiputti, Tavano, Vorano, De Luca, & Fabbro, 2002; Kiran & Edmonds, 2004; Marangolo, Rizzi, Peran, Piras, & Sabatini, 2009; Miertsch, Meisel, & Isel, 2009). Bilingualism has even been mentioned as a factor predisposing a person with a sensory aphasia toward the production of jargon (Perecman, 1989). Since processing routines linking conceptual with phonological form are assumed to be less automatized in bilinguals, Perecman and Brown (1981) suggest that phonological and semantic representations are decoupled from one another and jargon is, therefore, more likely in bilinguals than in monolinguals. So it can be expected that the less automated link between concepts and phonological forms is more suppressed in our patients which causes them to develop non-propositional (automatic) speech as a precursor of fluent aphasia (Wernicke/jargon aphasia).

These cognitive-linguistic considerations and interpretations of our findings in this single case of letter speech and our previous report on number speech must fit within a framework of neurophysiological alterations occurring in the context of acute stroke. All patients suffered from a major stroke involving a large part of the cortical and subcortical structures in the left middle cerebral artery territory. Although no attempt was made to assess lesion overlap in our patients, it seems unlikely that a strategic-focalized lesion of the cortex would entail this symptomatology and evolution. It is rather likely that these large lesions acutely cause a severe depression of cortical function, not only in the affected hemisphere, but most probably also in the contralateral hemisphere. From a physiological point of view, the acute onset of stroke with hypoperfusion is characterized by the occurrence of cortical spreading depression of electrophysiological activity and cortical spreading depolarizations, which are the cause of a number of cellular events leading to disturbance of ionic homeostasis, cytotoxic edema and disorders of water diffusion (Dreier, 2011). These acute events might be related to the hyperacute onset of mutism in our patients, although this is merely hypothetical. Similarly, in the very early stages of stroke a flaccid and complete paralysis is frequently observed, which spontaneously evolves over the course of hours or days into a milder motor dysfunction. Whether these clinical phenomena all reflect the same physiological events remains to be demonstrated, but it seems plausible to accept the idea of an acute "shock" of ipsilateral and perhaps also contralateral cerebral networks in the hyperacute stage of severe stroke.

Animal as well as human research indicate an increase of contralateral neuronal excitability within hours after stroke onset, which correlates to the concept of transhemispheric diaschisis and the well-known model of compensation attempts by activation of contralateral homologue cortical areas. It is tempting to link this contralateral activation to the onset of speech activity with an emphasis on content that is more dependent on right hemispheric networks, such as automatic overlearned sequences (Eagleman, 2009). Similarly, it is well known that emotionally driven content such as expletives recover more easily than propositional speech in some patients with aphasia (Van Lancker & Cummings, 1999). In the subsequent course of events, a gradual recovery as a function of compensation and plasticity occurs, leading to the onset of propositional speech, most probably reflecting activity within networks in the left hemisphere.

It remains unclear why not more patients with left hemispheric stroke develop this sequence of events and what makes the cases we have described unique. It seems that the presence of an initial stage of mutism, in the context of a fluent aphasia, with its characteristic pressure of speech, might entail a transient stage of category-specific utterances, consisting of items from automatic series or emotionally loaded speech utterances. These phenomena probably reflect underlying neurophysiological events and are correlated with severe stroke, involving a considerable part of the middle cerebral artery territory, and the development of compensatory or plastic contralateral hemisphere activity. This contralateral activity may be important in terms of prognosis and treatment decisions. So far, these cases with category-specific speech have evolved to jargon aphasia, with which it shares a number of characteristics as outlined above.

Disclosure statement

No potential conflict of interest was reported by the authors.

Ethics

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