APHASIOLOGY, 2012, i*First*, 1–16

Psychology Press
Taylor & Francis Group

Speaking in numbers as a transitional phase between mutism and Wernicke's aphasia: A report of three cases

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Background: Mutism in the context of hemispheric stroke with aphasia is rare and usually evolves to non-fluent aphasia.
Aims: We describe three multilingual and mathematically educated patients with an initial presentation of mutism, followed by a short-lasting episode of speaking in numbers as a transitional stage before developing Wernicke's aphasia. We discuss potential pathophysiological mechanisms underlying this phenomenon.
Methods & Procedures: Clinical neurolinguistic testing, including Aachen Aphasia Test and video-analysis of spontaneous speech by transcription.
Outcomes & Results: In the transitional stage between mutism and Wernicke's aphasia, numbers were randomly uttered in one or more languages, although not necessarily in the first acquired or most-used language. Number speech occurred not only in propositional speech, but also during reading and naming.
Conclusions: These patients exhibit two peculiar phenomena. First, the evolution of mutism to Wernicke's aphasia and second, the transitional phase of number speech. It is hypothesised that mutism may be more frequent in the hyperacute stages of stroke-related aphasia as a consequence of transient generalised failure of the language network. Current theories on the organisation of number magnitude and lexical retrieval of number and non-number words are discussed with reference to numerical speech. The lack of previous reports of this syndrome is probably due to the combination of the transient nature of the phenomenon and the suspected prerequisites of a specific neuro-anatomical lesion and educational background.

Keywords: Numbers; Mutism; Wernicke; Aphasia; Multilingual; Stroke.

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http://www.psypress.com/aphasiology http://dx.doi.org/10.1080/02687038.2012.660460

The literature on mutism in the context of classical left-hemispheric cortical stroke with aphasia is relatively scarce. The available data suggest that it most frequently occurs as a transient phenomenon, and usually evolves to a non-fluent type of aphasia. Non-fluent aphasia may further evolve into a fluent type of aphasia over the course of months or even longer (Pedersen, Vinter, & Olsen, 2004). The recent literature suggests that the presence of mutism in the initial stages adversely affects stroke prognosis (Oliveira & Damasceno, 2011).

In this report we will describe three multilingual and mathematically educated patients with a peculiar evolution after sustaining a classical left-hemispheric stroke. In the initial stages of stroke and following a transient state of muteness, all three demonstrated a short-lasting interval during which speech production was limited to items of one specific semantic category. More specifically they uttered only random sequences of numbers. After this intermediate stage, which lasted for only hours or days, they developed Wernicke's aphasia. These cases raise questions about the nature of Wernicke's aphasia and the neural organisation of number processing in aphasic patients.

The nosological position of Wernicke's aphasia has been the topic of debate since it was first described as a sensory aphasia by Carl Wernicke (1874). He defined this type of aphasia as a disorder of language in which comprehension is disturbed and pathology-specific jargon is produced. He attributed this to a lesion in the posterior parts of the superior temporal gyrus, which was subsequently called Wernicke's area. In the twentieth century a central issue in the discussion on what constitutes Wernicke's aphasia was the nature of the comprehension deficits. In agreement with Wernicke, Luria (1973) considered a phonological perception problem as the core problem. Blumstein, Baker, and Goodglass (1977), on the contrary, argued that a phonological problem ruled out the diagnosis of Wernicke's aphasia. She was followed by Brookshire (1997) who agreed on this and proposed a semantic problem as the core issue in the impairment of comprehension.

The discrepant views on the nature of the comprehension deficits can be interpreted as a consequence of the differences in the neuro-anatomical boundaries of the underlying lesions. The larger these lesions, the more neural substrates involved in linguistic processing will be included in this area. The neural substrates for phonological input are thought to be close to the auditory association areas in the left superior temporal lobe (Graves, Grabowski, Mehta, & Gupta, 2008). Studies on semantics have highlighted the implication of a large part of the temporal lobes (Binder, Desai, Graves, & Conant, 2009). The mid- to posterior superior temporal cortex is heavily involved in grammatical input processing (Friederici, Kotz, Scott, & Obleser, 2010). In addition a dynamic communication between both superior temporal lobes underlies the perception of segmental and suprasegmental features and is therefore essential for prosodic comprehension (Wildgruber, Ackermann, Kreifelts, & Ethofer, 2006). Tanner (2007) describes Wernicke's area not as a centre of auditory comprehension, but rather as a conduit for decoding of perceptual, phonological, grammatical, and semantic features of language. Consequently lesions in the area may cause different types of comprehension deficits, depending on the neuro-anatomy of the lesion.

In addition to comprehension problems, Wernicke (1874) also stressed the occurrence of expressive problems, which he called jargon. Recent neuro-anatomic models have stressed the network organisation of language compounds, connecting cerebral areas primarily activated in perception with the regions involved in language production (Dominey & Inuib, 2009; Hickok & Poeppel, 2007) and articulation (Hickok et al., 2007). A lesion in Wernicke's area, which is primarily associated with perceptual or comprehension problems, can also cause expressive dysfunction such as phonological or semantic jargon, paragrammatism, all or not combined with prosodic dysfunction. In addition, failing feedback systems will add to impaired speech. This means that Wernicke's aphasia, as a consequence of a lesion in Wernicke's area as defined in the broadest sense, may present with comprehensive and expressive disabilities in all language modalities.

Disturbances of number processing in aphasic patients have been reported before. Cipolotti, Butterworth, and Denes (1991) described a patient with a category-specific semantic impairment for numbers, in addition to impairments of other ordinal classes such as days of the week, months of the year, or reciting the alphabet, while overall semantic abilities were intact. Thioux et al. (1998) described a patient who had an inverse dissociation, with a relative preservation of numerical semantics in the context of a more generalised semantic impairment. Similarly Cohen, Verstichel, and DeHaene (1997) described a patient with neologistic jargon aphasia after a bi-hemispheric frontotemporal stroke, who displayed a category-specific advantage for the denomination of numbers.

Jefferies, Bateman, and Lambon Ralph (2005) reported a patient with semantic dementia, which is a temporal variant of frontotemporal lobe degeneration, who retained the notion of magnitude but lost the lexical access to numbers. These data suggest a network connecting parietal and temporal structures for semantic aspects of numbers. Consequently, brain lesions disrupting this network may interfere with knowledge and retrieval of numbers while other lesions, dependent on the neuro-anatomy, may affect numerical semantics less severely than other semantic categories. The current report, describing three patients with number names as the only utterances after mutism, contributes to the concept of the dissociable nature of the lexical retrieval of number words and non-number words.

To our knowledge this is the first paper reporting mutism followed by an episode of utterances reduced to one specific semantic category (numbers) in the initial stage of Wernicke's aphasia. The unanticipated nature of the observation and its short-lasting appearance made it impossible to perform specific neurophysiological or functional neuroimaging testing. A number of hypotheses underlying this phenomenon can be formulated, based on neuro-anatomic and neurophysiological models of language and number processing.

CASE DESCRIPTION

Case 1

A 53-year-old, right-handed, Dutch-speaking man (LW) was admitted to the hospital after an acute stroke. His personal medical history was unremarkable, as was his family history. During his professional career he had a leading function as an engineer in an automobile factory. This highly educated man was a native speaker of Dutch (L1), with an active knowledge of French (L2) and English (L3) and a passive knowledge of German (L4).

At the first neurological examination after admittance he was fully alert, but demonstrated mutism, evident comprehension impairment, and a right hemiparesis and hypoesthesia, involving more the upper than the lower limb.

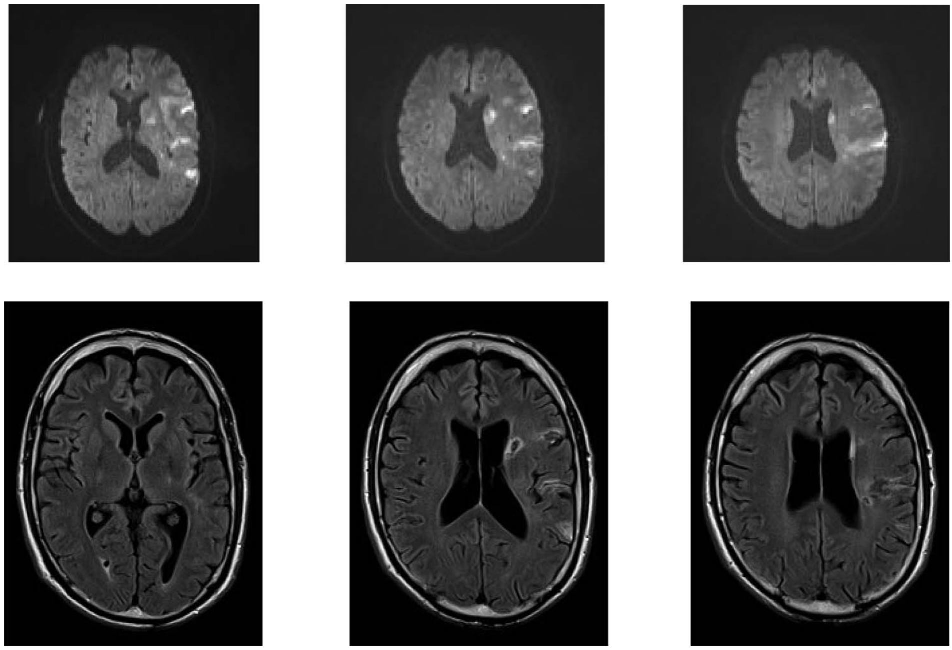

Figure 1. Diffusion-weighted MRI (upper row) in the acute stage of stroke and Fluid Attenuated Inversion Recovery MRI (lower row) at 6 months after stroke onset of patient 1. Diffuse ischaemia in the left middle cerebral artery territory involving the insula and the peri-insular cortex and the striatum.

MRI confirmed a lesion involving the left temporal, parietal and insular cortex, and left lentiform nucleus and caudate nucleus (Figure 1).

Linguistic status in the first hours after stroke. During the neurolinguistic evaluation LW presented with mutism and had severe difficulty in comprehending simple yes/no questions. His nonverbal communication behaviour was relatively spared, including eye contact, gesticulations, and head nodding. The patient was unable to repeat, to write or copy his name, or to read regular words. Finger agnosia, calculus, and orientation could not be evaluated because of the severe comprehension deficits.

Linguistic evolution. The mutism persisted for several hours. Suddenly the patient began to take initiative to speak, the first utterance being a number. That moment was the start of 2 days of speaking in numbers between 0 and 10, but 0 and 10 not included. These were expressed in Dutch (L1). Evaluations in other languages were not done during this period. The patient did not behave euphorically and had no logorrhea but on the contrary showed respect for turn taking in conversation. It appeared as if the patient attempted to speak fluently but was unable to find his words, compensating for his failure with the production of numbers at a slow rate. No formal confrontation naming test was taken during this short period of number speech, but the patient similarly used number words when naming objects in his room.

The following is a transcription of a video-recorded conversation between the patient and the speech language pathologist (SLP):

Patient [spontaneous speech]:	2, 2, 3, 2, 4, 2, 4, 2, 4, 5, 6, 2, 1, 2, 4, 2, 4
SLP:	*Are you married?*
Patient:	Yes
SLP:	*Do you have children?*
Patient:	7, 2 from 3 [he has one daughter]
SLP:	*How many children do you have?*
Patient:	1, 4, 1, second
SLP:	*What's your name?*
Patient:	1, 2, 2, 8

The numbers were pronounced in a randomised order, arythmically, without intonation, but with number and number group perseverations, preferring the number 2. Overviewing 30 utterances, the patient demonstrated five number perseverations and four number group perseverations.

On day three the numbers were replaced by function words and the conversation continued with the patient repeating the therapist's nouns and phrases. This echolalia was stopped by an intervention of the SLP. Subsequently the patient tried to speak fluently, but his speech was blocked by persistent word-finding problems. From that moment word reading resulted in phonological neologisms. Writing was possible but remained reduced to copying some letters. After a stable communication status for 1 week, the SLP decided to administer a standardised test (Akense Afasietest by Graetz, De Bleser, & Willmes, 1992) (see Table 1). At that moment spontaneous speech was slow and fluent with phonological paraphasias and neologisms. Repetitions were limited to neologisms and perseverations. Naming and comprehension were so disrupted that the tasks could not be performed. Reading was impossible and writing remained restricted to the copying of some letters. Nonverbal semantics as tested by the nonverbal semantic association test (Visch-Brink, Stronks, & Denes, 2005) were mildly impaired, resulting in a score of 21/30, with 6 errors of the second-degree relatedness. Acalculia was present at that moment.

After 6 months of intensive speech rehabilitation the patient had recovered almost completely (see Table 1). The Akense Afasietest could only demonstrate a few word-finding problems for infrequent words and some paralexias.

During his recovery this patient did not switch from one language to another. Although he was not encouraged to rehabilitate L2, L3, or L4, the patient reported on his follow-up consultation that these languages had the same recovery as L1.

TABLE 1
Results in Patient 1 on the Aachen Aphasia Test (Graetz et al., 1992) in the acute stage and at 6 months post onset

		Acute stage		*6 months post onset*	
Language modality	*Range*	*Score*	*Percentile*	*Score*	*Percentile*
Token Test	50–0	46	18	16	75
Repetition	0–150	25	9	147	97
Writing language	0–90	52	52	88	97
Naming	0–120	19	19	107	90
Language comprehension	0–120	0	1	117	99
		100% Wernicke's aphasia		80.7% amnestic aphasia 19.3% no aphasia	

Case 2

ISP, a right-handed, 70-year-old, multilingual man with a previous history of coronary artery bypass, was admitted in our department after being hospitalised in Switzerland in the context of an acute stroke with aphasia. He did not receive speech therapy during this week.

His native language was Hebrew, with French, English, and Dutch as L2, L3, and L4, respectively. Since he was living in the Dutch-speaking part of Belgium for more than 30 years, Dutch was his most-used language in daily communication. During his professional career he had been working as an engineer. This patient was transferred to our hospital 1 week after stroke onset.

Linguistic status in the first hours after stroke. ISP had remained mute since his stroke and had evident comprehension problems. Further neurological testing revealed no motor limb dysfunctions, or apraxia or visual field disturbances. His MRI demonstrated a large ischaemic lesion in the left middle cerebral artery territory, involving most of the perisylvian fronto-temporo-parietal cortex and the insula (Figure 2).

During the neurolinguistic screening he presented with mutism, showed no initiative to speak, and had severe comprehension problems. Even intensive speech stimulation could not help the patient to break through his mutism. Nonverbal communication was limited to eye contact. The patient was unable to assign objects or to match identical pictures on auditory command. On functional comprehension tasks (e.g., give me your hand, close your eyes, drink some water) he appeared to have the best comprehension in French (L3). Furthermore, the patient was not able to repeat, to write or copy his name, or to read regular words. Finger agnosia, calculus, and orientation could not be evaluated because of the strong comprehension deficits.

Linguistic evolution. The morning following his transfer the patient spontaneously started to speak and name pictures in randomly selected numbers, expressed in English. In response to questions with a predictable response, the spontaneous speech consisted of numerical series which were intonated, but the length of the series did not

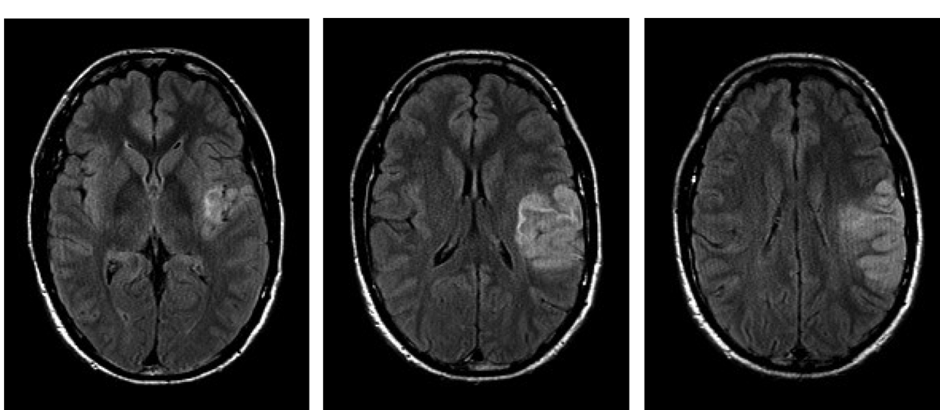

Figure 2. Fluid Attenuated Inversion Recovery MRI at 6 weeks after stroke onset in patient 2. Large ischaemic lesion in the insula and the adjacent temporo-parietal cortex of the left hemisphere.

equal or correlate with the word length or the number of syllables/words of the target sentences. From that moment on the patient's spontaneous speech was restricted to numbers, sometimes resulting in a logorrhoea of "number speech". The patient behaved in a frustrated and uncooperative way, probably due to a combination of a disturbed auditory feedback, anosognosia, and euphoria, which made it impossible to perform standardised testing. A few hours later his "number speech" was interrupted by intervals of "Polyglotte Reaktionen" (Minkowski, 1928) as in "Seven, douze, treize, do you understand me?", "Drie of vier, c'est vrai?" The following days the "number speech" disappeared more to the background and the "Polyglotte Reaktionen" and phonological neologisms increased, with a preference for English (L3), French (L2), and Hebrew (L1). He hardly produced Dutch words (L4), even though this had been the dominant language over the last 30 years. It appeared as if the patient used language switching or numerical speech as strategies to overcome his word-finding problems and to maintain his drive to keep the conversation going. When unable to activate target words, he replaced the words by random numbers:

The following is a transcription of a video fragment; French, *English*, [Hebrew], **Dutch**):

Patient: Cent douze, douze toize, seize, c'est comme ça *but* douze ans ils est, *but no, it is not, it is not,* non, c'est [Hebrew] *no*, pas c'est [Hebrew] *no,* **nu,** *I know yes, you* [Hebrew] *on differ,* cent dix ça et c'est deux, langue [Hebrew] lal [Hebrew]

SLP: C'est en hébreu alors?

Patient: Oui oui alors. Et *you want and perhaps you can?* Laisse *me seen what's,* non, *that is* maltraite. *I think it's all right*. Les qui a la même chose. Elle deux cents sept, cent dix deux deux mille.

SLP: Maintenant vous parlez en chiffres?

Patient: *No no it is nothing, but I want to note* elle deux ans d' habitude elle deux cents grand elle deux cents [Hebrew][Hebrew][Hebrew], *for this something is.* Deux cents cents deux, c'est quelque chose comme ça. **Ja, voor** *this, let me, you have this and this, you like this, for you to see.* Elle deux cents, elle deux cents grammes. C'est ça?

A transcription and analysis of a 200-word video-recorded spontaneous speech sample after the episode of isolated numerical speech confirmed a preference for L3 (80%), followed by L2 (11%), L1 (6%), and finally L4 (2%). A total of 18.5% of the words consisted of numbers, among which 91.9% were cardinals and 8.1% ordinals. We do not have a formal translation of the Hebrew utterances but, reportedly, at least some of them were also numbers. The mean length utterance (words) was 3.69, which was far below the Dutch norm of a mean of 8.22 (Wijckmans & Zwaga, 2005).

The spontaneous utterances contained 75.5% of empty utterances and 24.5% of syntactically, phonologically, and semantically correct utterances. The empty utterances sometimes consisted of perseverations (18.9%) with a preference for the French number two (deux), phonological paraphasias (1%), and neologisms (1%).

The patient hardly understood Dutch, which he had been using for at least 30 years during his stay in Flanders. Unfortunately speech therapy was unavailable in Hebrew (L1) in our institution and was therefore given in French (L2). Despite intensive therapy with a frequency of 1–2 hours/day, the patient's language recovered very slowly, and he was diagnosed with a severe Wernicke's aphasia with "Polyglotte Reaktionen". At the end of his hospitalisation (6 weeks after stroke) his spontaneous speech was fluent and trilingual (Enlish–French–Hebrew), empty, and full of word perseverations. His comprehension remained disturbed auditorily more than orthographically. In automatic series (days of the week) and confrontation naming he persisted in isolated numerical production until the end of his hospitalisation. In silent reading he could not match words with pictures. Reading aloud resulted in letter-by-letter reading, with a random number for every letter (for instance: lunettes (French word for glasses) = cent, douze, douze, trois, seize, douze, cent, dix). Writing was still limited to copying graphemes. Even writing graphemes was accompanied by oral number production. Interestingly the patient was unable to count from one to ten (for instance: 1, 2, 3, 4, 5, 6, were produced respectively as dix, third, two three, four, sixty). The frustrations resulting from the communication disturbances continued to prevent standardised testing. After he was discharged from the hospital the patient underwent an intensive period of speech rehabilitation at home, but without success. After moving to Israel 6 months post onset, he had a spectacular recovery in Hebrew comprehension and production (L1), which allowed communication in daily life again. According to his friend's report 1 year later, the pathological language switching disappeared in favour of his native Hebrew language while there was no recovery of the Dutch language. Unfortunately we were unable to retrieve formal linguistic test results of his recovered Hebrew. We have no information about the recovery of the English language.

Case 3

IST was a 60-year-old, trilingual—Bulgarian (L1), French (L2), Dutch (L3) and English (L4)—engineer who had been living in the French-speaking part of Belgium for 25 years as a political refugee. As a consequence of an acute stroke he was admitted to our hospital. Neurological examination revealed a right hemianopia without hemiparesis and MRI demonstrated an occipito-temporal intracerebral haemorrhage (Figure 3).

Linguistic status in the first hours after stroke. This patient was admitted to our hospital presenting with mutism and severe comprehension problems. The patient didn't

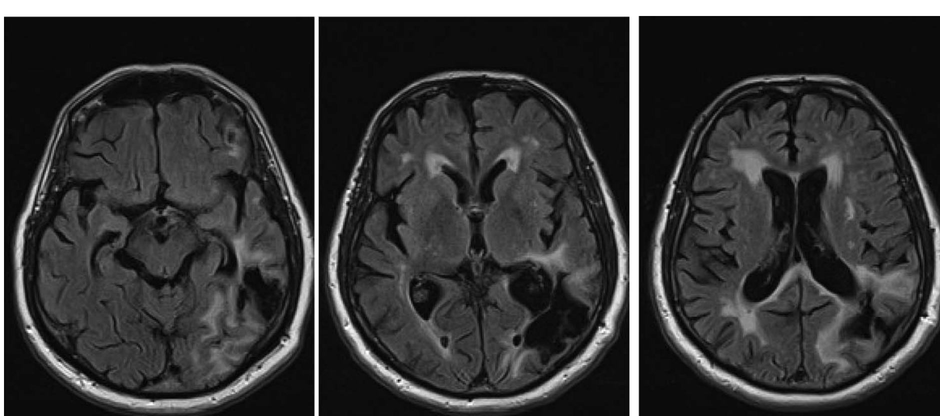

Figure 3. Fluid Attenuated Inversion Recovery MRI at 3 months after the acute hemorrhage in patient 3, demonstrating a large lesion in the temporal lobe with surrounding reactive tissue.

take the initiative to speak and was unable to assign objects or to match identical pictures on auditory command.

Linguistic evolution. The mutism was followed by a sudden episode of numerical speech in L2 which disappeared after a few hours. During this episode of numerical speech the patient was restless and rigid in his actions (e.g., he only wanted to eat sitting in the chair at the left side of the table with his book in front of him), which made standardised or bedside screening testing impossible. The "number speech" contained cardinal numbers from 1 to multiples of 10,000 and was produced with intonation, in a randomised order, and with a preference for the number 3. After some hours he spontaneously started to mix numbers with a fluently produced L2 (e.g., Nous avons vécu très, trois, toute ma vie), without attempting to correct himself. When asked to write down the languages in which he was proficient, he noted: "flamand 8000, français 80000, Bulg 500 and Russ 20 g". In the following hours and weeks a functional conversation became possible in a mixture of L2 and L3, but communication was hampered by logorrhoea, perseverations of stories and a tendency for empty speech, although his nonverbal communication remained intact. The oral production of the present patient was characterised by severe word-finding problems for frequent words and proper names, which were compensated for by phonological and semantic paraphasias. The phonological disturbances seemed equally severe in all language modalities. Remarkably the auditory feedback improved only slightly, resulting in a limited numbers of self-corrections. The patient seemed quite unaware of his speech errors. Orthography was disturbed on the level of orthographic discrimination and input lexicon. In contrast with the phonological output capacities, the patient could not rehearse an orthographic word form or fill in pre-selected graphemes in visually presented word forms matched to pictures. As in the other cases described above, the comprehension problems and agitated behaviour prevented standardised testing or bedside screening or evaluation of calculia, finger agnosia, and orientation. After 8 weeks of hospitalisation and intensive speech treatment in French (5 days a week, 1 hour daily) the patient's spontaneous speech remained fluent with a preference for L2. He was able to perform simple commands given in French and could orthographically complete simple sentences in L2 with minimal phonological paragraphias. Calculation was intact. After 2 years of intensive rehabilitation in French the patient consulted the SLP (MDL) in Dutch with some "Polyglotte Reaktionen" in French that the patient did not correct. Standardised testing (Akense Afasietest) was hardly possible due to the frustrated and agitated behaviour of the patient. The token test resulted in a score of 31/50, repeating of sounds in 18/30, repeating of mono- and polysyllabic words in 56/90, and repeating of sentences in 8/30. Reading aloud resulted in 20/30. This task as well as the remaining tests had to be interrupted because of too many frustrations.

DISCUSSION

This paper reports three stroke patients who developed a relatively short-lasting "number speaking" as a transition between mutism and Wernicke's aphasia. These cases are peculiar for two reasons. First, it is remarkable that patients evolve from a state of mutism to Wernicke's aphasia over such a short period. Second, the transitional phase with number speech is a phenomenon that, to our knowledge, has not been described before.

Evolution to Wernicke's aphasia

Mutism in the acute stage of aphasia can probably be considered an extreme form of expressive dysfunction as a consequence of a kind of "paralysis of the language network" or diaschisis. Assuming this possibility, it could be hypothesised that mutism occurs more frequently in the hyperacute stages of different types of stroke-related aphasia. However, this hypothesis needs to be confirmed in larger prospective series including linguistic evaluation in the earliest stroke stages, which is not always easily obtained.

Number speech

Our three cases had a transient stage of number speech with a number of similarities. In all three patients the numerals were expressed randomly. Sometimes there was a tendency to perseveration of one or two numbers with preference for one number (patients 1 and 2: preference for number 2; patient 3: preference for number 3). In none of the cases was the ordinal sequence respected. The randomised number production excludes the hypothesis of automatic speech or overlearned rote verbal material, as one should expect that patients would start to count or produce other series (Cohen et al., 1997). Remarkably, the patients share the same professional (engineering) and multi-lingual education (with French and English as their second and third acquired language respectively). To our knowledge this is the first report of the phenomenon of numerical speech, more specifically in propositional speech, naming, and reading, as a precursor of Wernicke's aphasia.

The underlying aetiology of this type of numerical speech can be discussed on different but related levels of number specificity, network balance, and neuro-anatomic connections.

Number specificity. The isolated numerical utterances could be considered a form of "category-specific recurrent utterances" (RU). RU is a term that was introduced by Hughlings-Jackson (1879), but throughout history it has received different names such as monophasia, verbal stereotypy, or speech automatism. RUs are variably defined, but probably the most comprehensive description of the phenomenon is that of Huber, Klingenberg, Poeck, and Willmes (1983) who defined it as recurring, stereotyped utterances that consist of neologistic sequences, words, or phrases that neither lexically nor syntactically fit into context and are produced against the presumed intention of the patient. RUs can consist of single syllables, strings of syllables, words or word strings, sentences, or neologistic utterances. The RU is the only form of speech output or dominates verbal communication, although rare interjections of adequate speech may occur in specific social or emotional situations, which Jackson referred to as "occasional utterances". Moreover, in some patients the RUs display a more varied presentation with phonologically related forms or phonological variations of a stereotypical core form (Blanken, Wallesch, & Papagno, 1990). RUs can persist for variable periods of time after onset, ranging from hours to even years.

Although it has been acknowledged that RU can occur in all clinical types of aphasia (Basso, 2004), series of patients with RU contain only few if any patients with Wernicke's aphasia (27 patients, 1 Wernicke aphasia in Blanken et al., 1990). Moreover, the original view that persisted for a large part of the twentieth century was that patients with RU had a normal understanding of language (Broca, 1861; Hughlings-Jackson, 1879). RUs may be pronounced with or without intonation, but the added communicative value of such prosodic variations to the already poor content of the verbal utterances seems to be limited.

Different types of RU have been described. Hughlings-Jackson (1879) distinguished incomprehensible jargon utterances, real words, phrases, and yes/no particles. Code (1982a) differentiated two classes of RU: real word recurring utterances (RWRU), mostly consisting of words or word strings, and non-meaningful recurring utterances (NMRU), most frequently consisting of syllables with consonant-vowel structure or vice versa. Blanken and Marini (1997) preferred the terminology of lexical and non-lexical automatisms.

Hughlings-Jackson (1879) suggested that the RU simply reflected the fragment that the patient was speaking at the time he or she was struck by the brain lesion. He considered the patient to be blocked in speech so that it was impossible to move to a next sequence. Conversely, a number of authors have suggested that the RU reflects the first segment of speech after the onset of aphasia (Code, 1982b). This of course does not explain the full range of possible phenomena described above, such as neologistic jargon, strings of syllables, or the occurrence of more than a single RU in one patient. A popular hypothesis on the aetiology of RU is the two-source hypothesis, which proposes different sources for RWRU and NMRU (Code, 1982a; Code, 1994; Wallesch & Blanken, 2000). RWRU are proposed to be generated by an intact right hemisphere. These RU are often elements of automatic series, proper names, frequent words, or emotionally charged utterances, which are supposed to have a non-propositional origin. NMRU, on the contrary, are suggested to originate from the damaged left hemisphere which results in neologisms or the generation of syllables, but rather as propositional speech. Despite its attractiveness, the two-source hypothesis cannot explain all characteristics of RU. It is difficult to reconcile the hypothesis with the occurrence of infrequent or non-emotional words as RWRU. In addition the phonological variability of RU sometimes results in a mixture of RWRU and NMRU, which indicates a common source. More fundamentally this hypothesis does not explain the limited number of verbal utterances, which is the most important characteristic of RU. Wallesch and Blanken (2000) therefore argued that lexical and non-lexical automatisms are generated by a similar mechanism, but in addition they advocate a separation of mechanisms of initial utterances, in which limbic or right-hemispheric systems may play an important role, from mechanisms subserving the continuous and stereotypical production. A possible explanation for the latter is a pathological blocking of the articulatory buffer systems (Blanken et al., 1990). An attractive hypothesis for the perseverative nature of utterances was offered by Cohen and Dehaene (1998), who suggested perseverations as a consequence of a lack of input in a given processing level, which causes a persistent activity that stems from previous trials and that is no longer overcome by a current or new input. As such, they relate the occurrence of perseverative utterances to the mechanisms involved in priming. When taking into account the fact that these mechanisms can apply to all language modalities, one can assume that priming mechanisms may be involved in the generation of stereotypic, perseverative utterances at the level of sounds, phonemes, words, or word strings, or at the level of discourse. Martin and Dell (2004) largely corroborated this hypothesis, but argued the importance of a number of variables. First, word frequency must be taken into account. If familiarity of the upcoming item is low, the probability of switching to this less-familiar item will be lower. Conversely, one can assume that if semantic or phonological relatedness is higher, then the probability of remaining within the same semantic or phonological category could be higher. A second variable is speech rate. If there is little time to switch to an upcoming item, the odds of repeating the previous item or sticking to phonologically or semantically related items may be increased. A third variable is the obvious nature and extent of the brain damage, leading to a change in the weight of connections between different processing levels in the different linguistic modalities.

To a large extent the utterances of number speech in our patients fit the description of Huber et al. (1983), although they lack the typical recurring, stereotypic nature of classical RUs. It could be argued that in this case the recurring and stereotypical nature does not apply to the utterances in themselves, but rather to the retrieval of items from a single semantic category, the numerical category more specifically. Previous reports in the literature have indicated other category-specific utterances such as curses. In addition to the early reports of Broca (1861) and Hughlings-Jackson (1879), more recent investigations by Code (1982a) have indicated that swear words and expressions frequently occur as RU. Different from numerical speech, curses are probably more emotionally mediated by limbic resources. However, this does not explain the category specificity of these utterances. A plausible hypothesis is the one raised by Cohen and Dehaene (1998) and further elaborated by Martin and Dell (2004) concerning the pathophysiology of perseverations. The authors suggest a persistent activation of retrieval processes within a specific semantic class (e.g., numbers), if the activation of moving to another category is deficient, or alternatively if disengagement from the current category is persistently inhibited by the brain damage.

Network balance. As outlined above, neuronal damage may lead to a change in the weights of connections between different levels of processing. Therefore, in the first stage after stroke before plastic changes intervene, semantic networks may operate in an unbalanced way (Martin & Dell, 2007). In addition, it is assumed that the most frequently used networks during life offer the most resistance to brain damage (Butterworth, Cappelletti, & Kopelman, 2001). The number class is hypothesised to be highly frequent and overlearned in our three mathematically educated patients (engineers). As far as we know, no other cases of category-specific utterances related to profession or training have been reported in the literature. As such, the preservation of one semantic class could be considered as equivalent to the preservation of the first acquired (law of Ribot) or most frequently used (law of Pitres) language after stroke.

Neuro-anatomic connections. Numbers probably constitute a specific semantic category, with a separate neuro-anatomic and neurophysiologic organisation. Dehaene, Piazza, Pinel, and Cohen (2003) propose a tripartite organisation of number-related processes in the parietal lobe. The horizontal segment of the intraparietal sulcus appears to be the core quantity system, supplemented by two other circuits: (1) a left angular gyrus area, in connection with other left hemispheric perisylvian areas, which supports the manipulation of numbers in verbal form, and (2) a bilateral posterior superior parietal system, which supports attentional orientation on the mental number line. We wonder if this bilateral representation "ensures" that digital information is less vulnerable to unilateral brain lesions. Recently a voxel-based lesion study revealed that word reading on the one hand and Arabic number and number word reading on the other hand rely on two distinct brain networks. Whereas a frontal network, including Broca's area and the premotor cortex, is involved in word reading, Arabic number and number word reading seem to rely on temporo-parietal brain regions (Piras & Marongolo, 2009).

These findings support the previous observations on number processing in aphasia mentioned in the introduction.

Multilinguality and feedback. Remarkably the three patients made no attempt to correct their number speaking in spontaneous speech, nor did they remember it afterwards, which can be explained by the localisation of their lesion. The language-dominant temporal plane, which is considered as an integration centre for auditory and somatosensory feedback (Dhanjal, Handunnetthi, Patel, & Wise, 2008) and which is driven predominantly by feedforward predictive corollary discharges (Paus, Perry, Zatorre, Worsley, & Evans, 1996), is involved in the brain lesion in all three patients. We hypothesise that the lesion of the temporal plane in these patients caused a suppression of the connection between the higher-order left ventrotemporal and the inferior frontal neocortex in the first few hours, resulting in a temporary dysfunction of the auditory feedback system, which compromises the cognitive control of language and hence the ability for self-correction. This explains why these three patients did not correct themselves in their numerical speech nor in their language switching after the numerical speech period. Moreover, this hypothesis is strengthened by studies in low- and high-proficient bilingual people, in which the temporal plane (together with the pars triangularis and pars opercularis) and the left inferior frontal cortex (together with the caudate and the pre-supplementary motor area/anterior cingulate) have been proposed as the cortical areas involved in language switching (Garbin et al., 2011; Parker-Jones et al., 2011). The transition from the episode of language switching to the selection and use of a single language in the three patients could be explained by two phenomena: (1) a functional recovery of the temporal plane itself or (2) a functional recovery of the cognitive control network connecting the temporal plane and the inferior frontal area.

(1) The transition from numerical speech to oral language production, with the ability to switch to other categories than numbers, in patients 1 and 3 could thus be considered as a recovery of auditory feedback, which was not the case in patient 2. The partial recovery of languages in propositional speech in contrast to the persistence of number speaking in non-propositional speech in patient 2 could then be interpreted as an improvement in the functions of the temporal plane, facilitating the selection of more than one frequent category (numbers), but still limited to the most frequent and stereotypic words and short sentences of all recovering languages (L1, L2, and L3).

(2) A functional recovery of the neural activity in the cognitive control network connecting the temporal plane and inferior frontal area, facilitates in bilingual people the recovery of the control of interference from words outside the target language (Rodriguez-Fornells, Rotte, Heinze, Nösselt, & Münte, 2002). Indeed, in the literature on bilingual language processing there is now a consensus that representations from both languages are always active (Van Assche, Duyck, Hartsuiker, & Diependaele, 2009), so that bilingual speech requires a cognitive control mechanism to handle this constant competition between languages. These processes are under control of the basal ganglia and frontal lobe circuits, which are also responsible for nonverbal cognitive control (Abutalebi, Rosa, Tettamanti, Green, & Cappa, 2009). Ansaldo, Marcotte, Scherer, and Raboyeau (2008) reported a patient with damage to these subcortical (left basal ganglia) circuits, who showed pathological language intermixing as a result. This is also observed here in patients 2 and 3. Also, similar to our patient 3 who

initially showed more loss for the first-acquired language (L1), Adrover-Roig et al. (2011) also reported a Basque–Spanish bilingual who suffered L1 production deficits and executive functioning deficits, after a lesion to the left basal ganglia.

GENERAL CONCLUSIONS

In conclusion, this is the first manuscript reporting "number speaking" as a brief transition episode, lasting only for hours, between an initial phase of mutism and subsequent Wernicke's aphasia. We assume that, in order to develop the phenomenon of numerical speech, both the specific neuro-anatomy of the lesion and the prior mathematical education of the patients are necessary prerequisites. The specific constellation of this syndrome, in combination with the transient nature of this phenomenon and the alertness from the caregivers for this kind of speech, could explain why this syndrome has never been described before. To illustrate its rarity we have only seen this syndrome in the three patients described in this report who were admitted to our hospital over a period of more than 10 years. Alertness for this phenomenon in future patients might allow examination of numerical speech in monolingual patients and evaluation of our hypotheses on the underlying pathophysiology with functional neuroimaging. The transient nature of the syndrome raises the question of a possible influence of number speech on the recovery and prognosis of aphasia.

Manuscript received 2 November 2011
Manuscript accepted 17 January 2012
First published online 29 February 2012

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