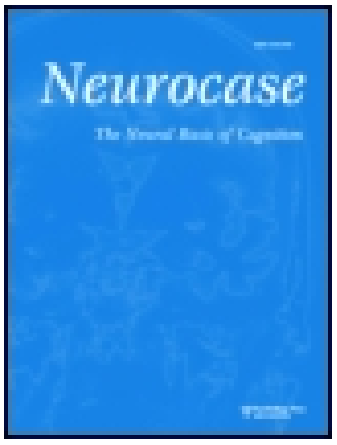

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Transient paligraphia associated with severe palilalia and stuttering: A single case report

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Transient Paligraphia Associated with Severe Palilalia and Stuttering: A Single Case Report

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Abstract

We report a patient who presented with a head injury associated with a brief loss of consciousness. Oral and written language characteristics were analysed on two different occasions. Twenty-five months after the head injury, some mild naming difficulties were found, associated with a notable dysfluency, corresponding to a mixture of stuttering and palilalia. In addition to his speech dysfluency, paligraphia, a tendency to word iterations when writing, was observed. Forty-five months after the head injury, severe palilalia and stuttering were still found, but paligraphia had disappeared. Palilalia and stuttering were severe in spontaneous language. No dysfluency was found in confrontation naming and verbal fluency tests. In language repetition, a minimal number of iterations was observed.

Introduction

Palilalia is a relatively unusual speech disorder characterized by involuntary reiterative repetition of words or phrases during verbal output (Benson and Ardila, 1996). Traditionally, palilalia has been thought to indicate bilateral basal ganglia involvement (Brain, 1961; Ackermann *et al.*, 1989). Palilalia has also been reported in patients with paramedian thalamic damage (Yasuda *et al.*, 1990), Parkinson's disease (Ackermann *et al.*, 1989), bilateral stereotaxic thalamotomy (Stracciari *et al.*, 1993), thalamic infarcts (Abe *et al.*, 1993; Casado-Chocan *et al.*, 1995), Tourette syndrome (Comings and Comings, 1987; Cardoso *et al.*, 1996), progressive supranuclear palsy (Uyama *et al.*, 1989; Kluin *et al.*, 1993), following right hemisphere damage (Horner and Massey, 1983), in recovery from severe aphasia, and in later stages of degenerative brain diseases (Hier *et al.*, 1985; Cummings *et al.*, 1988; Kashii *et al.*, 1990). Paroxysmal palilalia has been observed in association with epilepsy (Ardila and López, 1988). Neuroleptic-induced palilalia has also been reported (Garcia, 1990). Bilateral frontoparietal hypoactivity has been demonstrated by SPECT during transient palilalia (Dierckx *et al.*, 1991).

The literature on palilalia does not mention similar iterative errors in words or phrases when writing. In fact, reiterative writing, paligraphia, might indicate that the disorder is not limited to motor speech output and a broader motor language expressive disorder would deserve consideration.

The literature is limited for acquired stuttering. Acquired stuttering has been defined as a disorder that affects speech rhythms, making articulation of desired speech difficult due to involuntary repetitive prolongation or cessation of sounds (phonemes and syllables) (Nass *et al.*, 1994). Different conditions have been related to acquired stuttering. Fleet and Heilman (1985) ascribe acquired stuttering of gradual onset as due to degenerative diseases, such as Parkinson's or Alzheimer's disease. They describe the acute cases as being brought on by head trauma, emotional stress or stroke. Bhatnagar and Andy (1989) point out that developmental stuttering and acquired stuttering differ by their pattern; speech blocks are not confined to syllables and initial sounds in acquired stuttering. There are also no secondary symptoms with acquired stuttering, such as facial contortions, anxiety or struggles with articulation.

Stuttering and palilalia are different, even though they may be presented together. Acquired stuttering is more often repeating initial sounds (phonemes) and syllables, while palilalics usually repeat words and phrases at the end of an utterance. Acquired stutters tend to repeat all types of words and speak with normal melody in their voice, whereas palilalics repeat words with greater intrinsic meaning and they speak with amelodic tone (Fleet and Heilman, 1985).

It appears that acquired stuttering is most often attributed to left-sided or bilateral lesions. The anatomical structures involved in acquired stuttering are primarily cortical substrates (Bhatnagar and Andy, 1989). Lesion sites that have been associated with stuttering are the corpus striatum, the dominant parietal and temporal lobes, and Broca's

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area, including the immediate surroundings (Fleet and Heilman, 1985). It is unusual to find acquired stuttering after right hemisphere damage. However, Ardila and López (1986) found right hemisphere damage suggesting a tendency for repetitions of phonemes and syllables. Acquired stuttering may also be accompanied by aphasia (Fleet and Heilman, 1985; Ackermann *et al.*, 1996). Increases in repetitions have been found in Broca, conduction, anomic and Wernicke aphasia, as well as transcortical motor aphasia (Ackermann *et al.*, 1996). The most characteristic feature of acquired stuttering appears to be repetition of words in any position (Ackermann *et al.*, 1996).

Here we report the unusual case of a patient with a head injury, who presented with mild language difficulties, stuttering, palilalia and transient paligraphia.

Case report

A 55-year-old right-handed man presented with a history of head injury in a bicycle accident. The patient was a physician with speech and language within normal limits until the accident. He was unresponsive at the scene with a loss of consciousness for approximately 30 min and a post-traumatic amnesia of approximately 3 h duration. The Glasgow Coma Scale at the injury site was estimated to be 9.

The patient suffered severe lacerations and abrasions over the face, requiring plastic surgery. During hospitalization, CT scans, an MRI of the brain and an X-ray of the skull were interpreted as normal.

Four days post-trauma, the patient developed photopsia in his right eye, diagnosed as a retinal tear and posterior vitreous detachment. He was subsequently found to have a moderate aphasia evidenced by paraphasias and word-finding difficulties. Language was circumstantial and tangential with notable pressure of speech. Palilalia was observed during oral speech at this time. Cognitively, the patient evidenced significant perseveration and difficulty sustaining attention. Two months after the injury, his Full Scale IQ (Wechsler, 1974) was estimated to be 104 (61st percentile).

Neurological evaluation 3 weeks after the head injury revealed a gaze-evoked nystagmus and a truncal ataxia noted on heel to shin testing. EEG noted scattered theta transients bihemispherically. He was on no medication.

As the aphasia cleared, dysfluency became more pronounced with a speech pattern that showed palilalia, stuttering and semantic errors.

First follow-up evaluation

Twenty-five months after the head injury, his Verbal IQ was estimated at 114 (82nd percentile), his Performance IQ was 105 (63rd percentile) and his Full Scale IQ (Wechsler, 1974) was 110 (75th percentile). His performance on the Logical Memory (immediate and delayed recall) of the Wechsler Memory Scale-Revised (Wechsler, 1986) was equivalent to the 24th and fifth percentile, respectively. Non-verbal memory was superior to verbal memory (35th percentile). No evidence of ideomotor apraxia was found. Constructional tests revealed significant difficulties in organization and planning. The Rey–Osterrieth Complex Figure (Osterrieth, 1944) copy score was below the first percentile. Finger Tapping (Reitan and Wolfson, 1985) performance was normal. Fine manual dexterity and speed were normal on the left hand (76th percentile), but reduced on the right hand (27th percentile).

Table 1. Iterations observed in a 207-word spontaneous language sample (first follow-up evaluation)

	Number of iterations
Syllables	13
Words	95
Two words	3

At this time, some mild naming difficulties (anomia) were found, associated with a notable dysfluency, corresponding to a mixture of stuttering and palilalia. Confrontation naming ability, as measured by the Boston Naming Test (Kaplan *et al.*, 1978), fell between the second and fifth percentile. Word fluency, as measured by the Controlled Oral Word Association Test (Benton and Hamsher, 1976), was equivalent to the 77th percentile, but was characterized by perseverative responses. Performance on the WAIS-R Vocabulary subtest was equivalent to the 31st percentile. Performance on the Wisconsin Card Sorting Test (Heaton, 1981) was within the normal range. His dysfluency could be described as mixed stuttering and palilalia. Table 1 presents the number of syllables, words and phrase iterations found in a 207-word sample.

In addition to the speech dysfluency, a remarkable tendency of word, and even phrase, iterations was present in his writing. Written iterations first observed during the initial language evaluation continued to be observed presently even though the patient was aware of the deficit. In different written samples including 652 words, 13 word repetitions were found. Most of the repetitions were observed in short words. Figure 1 presents some examples of the patient's written iterations.

The patient was aware of these naming and fluency deficits, and he spontaneously referred to them and analysed them with some detail. The patient denied the use of any substance potentially responsible for his speech defects.

Positron emission tomography taken 35 months after the traumatic head injury was reported as abnormal. Decreases metabolically in frontal pole, dorsolateral prefrontal cortex, parietal cortex, subcortically in thalamus and putamen, temporal tip, orbitofrontal cortex, hippocampus, cerebellum, insular cortex and left visual cortex were observed. This pattern of abnormalities was considered compatible with diffuse brain injury.

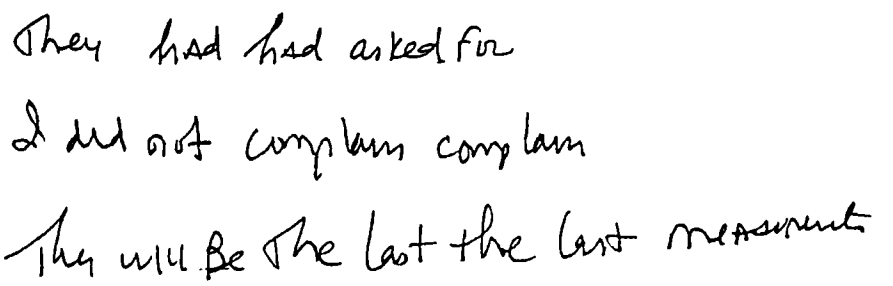

Fig. 1. Three examples of paligraphia.

Second follow-up evaluation

A new follow-up evaluation 45 months after the head injury revealed improvement in his naming deficit, but not evident improvement in his speech deficit. Confrontation naming ability, as measured by the Boston Naming Test (Kaplan *et al.*, 1978), corresponded to the 25th percentile. Word fluency, as measured by the Controlled Oral Word Association Test (FAS), was equivalent to the 40th percentile. Only two perseverative responses were observed. Semantic verbal fluency (to name as many animals as possible in 1 min) corresponded to the 10th percentile. Repetition subtest from the Multilingual Aphasia Examination (Benton and Hamsher, 1976) corresponded to the 15th percentile. No language comprehension deficits were observed in the Token Test (De Renzi and Faglioni, 1978) (50th percentile).

Stuttering and palilalia were observed with a similar degree of severity. Paligraphia, however, was no longer observed either in spontaneous writing (written description of Plate #1 from the Boston Diagnostic Aphasia Examination = 108 words; written description of his accident = 157 words) or writing to dictation (80 words—high and low frequency, short and long; and 10 sentences to dictation). No spelling or grammar errors were noted in the writing sample.

In order to pinpoint the characteristics of stuttering and palilalia, five different types of language conditions were used.

1. Spontaneous language: oral description of Plate #1 from the Boston Diagnostic Aphasia Examination (Goodglass *et al.*, 1983).
2. Spontaneous language: conversational language. The oral description of the patient's accident was recorded for further analysis.
3. Repetition subtest from the Multilingual Aphasia Examination (Benton and Hamsher, 1976).
4. Verbal fluency: a phonological condition (to produce words beginning with the letter F, A and S) and a semantic condition (to produce words corresponding to animals and fruits).
5. Confrontation naming: Boston Naming Test.

Palilalia and stuttering were only observed in spontaneous speech and repetition. However, dysfluency was notoriously higher in spontaneous than in language repetition. No dysfluency was observed in confrontation naming and verbal fluency tests. Table 2 presents the results obtained. Stuttering and palilalia are quite severe in spontaneous language, and virtually absent in the other language condition, except one-word and one-sentence iteration in language repetition. Seventy-six percent of the iterations are words, two-word and phrase repetitions, whereas phoneme and syllable iterations correspond to only 24% of the total number of iterations. In consequence, palilalia clearly predominated over stuttering.

Interestingly, it was noted that in dysfluencies, the iterated word (or phoneme, or syllable, or sentence) was frequently repeated several, even many, times. In consequence, the number of times the phonemes, syllables, words, two words and phrases were repeated in spontaneous language was analysed (Table 3). It is observed that, most frequently, a verbal element was repeated about 2–5 times, but, occasionally, the patient repeats the same phoneme or word over 15 times.

Finally, iterations in different sentence elements were analysed: nouns, articles, prepositions, adjectives, verbs and others (Table 4). Evidently, dysfluency particularly involved prepositions (13%) and 'others' (pronouns, conjunctions and adverbs) (21%). Iterations in adjectives was lowest (1%).

Discussion

In the present case, a severe palilalia was associated with transient paligraphia. Unfortunately, it is difficult to conclude from previously published cases whether paligraphia is or is not frequently associated with palilalia. If palilalia is often associated with paligraphia, the iterative defect would appear to represent a broader expressive language disorder, not simply a peripheral speech defect.

It must be emphasized that, in the current case, palilalia was considerably more severe than paligraphia. As a matter of fact, paligraphia was mild. In a written sample of 167 words taken 25 months after the head injury, only three word repetitions were found, whereas in an oral sample of 207 words, 21 iterations were observed. Twenty months later, no paligraphia was found under different writing conditions.

Table 2. Number of iterations observed in different language conditions (second follow-up evaluation)

	Spontaneous language		Repetition (280 words) (28 sentences)	Verbal fluency (60 words)	Naming (60 words)
	Picture description (8 sentences)	Conversational language (102 sentences)			
Phonemes	1	8	0	0	0
Syllables	2	10	0	0	0
Words	10	42	1	0	0
Two words	0	8	0	0	0
Phrases	0	4	1	0	0

In spontaneous language, 840 words corresponding to 110 sentences were analysed.
Stuttering occurred in 79/110 sentences.

Table 3. Number of times phonemes, syllables, words, two words, and phrases are repeated in spontaneous language (second follow-up evaluation)

	2	3	4	5–6	7–8	9–10	10–15	>15
Phonemes	4	0	0	0	1	0	2	2
Syllables	5	2	3	1	0	0	1	0
Words	16	8	2	14	4	0	1	7
Two words	3	2	1	0	1	0	1	0
Phrases	0	2	1	1	0	0	0	0

A total of 840 words corresponding to 110 sentences were analysed.

Table 4. Iterations observed in different sentence elements (second follow-up evaluation)

	Nouns	Articles	Prepositions	Adjectives	Verbs	Other
Picture description (103 words)	5/33	1/16	3/16	0/1	1/20	3/17
Percentage	15%	6%	19%	0%	5%	18%
Conversational language (737 words)	21/281	5/61	8/62	1/85	12/189	13/59
Percentage	7%	8%	13%	1%	6%	22%
Total (840 words)	26/314	6/77	11/78	1/86	13/209	16/76
Percentage	8%	8%	14%	1%	6%	21%

A total of 840 words corresponding to 110 sentences were analysed.

Improvement in speech was not observed in the follow-up evaluation performed about 4 years after the head injury. His speech deficits remained unchanged. Stuttering (phoneme and syllable iterations), and especially palilalia (word and phrase iterations), were severe. About a quarter of his dysfluencies corresponded to stuttering and three-quarters to palilalia.

Interestingly, dysfluency was strongly task dependent. Dysfluency was restricted almost completely to spontaneous language. In verbal fluency and naming tests, no iterations were found. It is not easy to understand such a significant dissociation in dysfluency, depending upon the linguistic context. It may be conjectured, however, that verbal fluency and naming are both tests tapping lexical knowledge. Lexical knowledge has been associated with retro-rolandic brain areas, very specifically left temporal areas (Benson and Ardila, 1996). Spontaneous language, on the other hand, requires the creation of active language, the assembling of words in a sentence, the expression of ideas and intentions in a verbal output. This creative verbal activity is assumed to be mediated by the left frontal brain areas (Luria, 1976).

Notably in our case, the patient repeated the very same verbal element many times, even over 15 times. To the best of our knowledge, this is extremely infrequent, in stuttering and in palilalia. This extreme iteration was evident in language sounds (stuttering) and in words (palilalia).

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Abstract

We report a patient who presented with a head injury associated with a brief loss of consciousness. Oral and written language characteristics were analysed on two different occasions. Twenty-five months after the head injury, some mild naming difficulties were found, associated with a notable dysfluency, corresponding to a mixture of stuttering and palilalia. In addition to his speech dysfluency, paligraphia, a tendency to word iterations when writing, was observed. Forty-five months after the head injury, severe palilalia and stuttering were still found, but paligraphia had disappeared. Palilalia and stuttering were severe in spontaneous language. No dysfluency was found in confrontation naming and verbal fluency tests. In language repetition, a minimal number of iterations was observed.

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O167

Primary diagnosis of interest

Paligraphia

Author's designation of case

The patient

Key theoretical issue

- Speech and motor disorders

Key words: paligraphia; palilalia; acquired stuttering

Scan, EEG and related measures

CT scans and MRI normal; PET: hypometabolism in several brain areas

Standardized assessment

Neurological examination

Other assessment

Language and speech assessment

Lesion location

- Diffuse brain injury

Lesion type

Head injury

Language

English