

The Impact of Left Temporal Lobe Glioma on Inner Speech

Vanja Kljajevic^{1,2*}, Estibaliz Ugarte Gómez³, Johanne Hatteland Somme⁴ and Cristina López³

¹University of the Basque Country (UPV/EHU), Vitoria, Spain

²Ikerbasque, Basque Foundation for Science, Bilbao, Spain

³Asociación de Traumatismo Cráneo-Encefálico y Daño Cerebral Adquirido (ATeCe) Alava, Vitoria, Spain

⁴University Hospital Alava, Vitoria, Spain

*Corresponding author: Vanja Kljajevic, University of the Basque Country (UPV/EHU), Vitoria, Spain, E-mail: vanja.kljajevic@gmail.com

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Abstract

Background: Inner speech has an important role in many human cognitive functions. Even though it has been extensively studied in post-stroke aphasic patients, there is currently no understanding on how slow-growing lesions affect inner speech abilities.

Methods: In the present study we investigated inner speech abilities in a 41-year-old, right-handed man, MN, who was diagnosed with low-grade glioma three years earlier. We tested his performance on a wide range of standard cognitive tests and on four experiments specifically designed to assess inner speech abilities, involving: silent rhyme judgments, syllable discrimination, and identification of words in compounds and names for numbers. Control data were obtained from 10 neurologically intact adults.

Results: The modified t-test revealed that, in comparison with the HC group, MN's silent rhyming was considerably worse ($p=0.027$), but his performance on the remaining tests was spared (overt rhyming: $p=0.136$; words in compounds: $p=0.288$, discerning syllables: $p=0.268$; discerning words in names for numbers: $p=0.48$).

Discussion: Overall, our data on inner speech in this particular case of left temporal lobe slow-growing tumor supports the notion that the brain retains considerable potential for functional reorganization. MN's performance on silent rhyming is likely due to a word retrieval deficit (since we used a pictorial paradigm in this task) and exacerbated by verbal working memory deficit.

Conclusion: Given the critical role of inner speech in various mental functions, assessment of this ability in patients with left temporal lobe tumors is highly recommended.

Keywords: Inner speech; Word retrieval; Anomia; Low-grade glioma; Left temporal lobe; Neuroplasticity

Introduction

Inner speech represents an important component of human mental life. It is often defined as silently talking to oneself or speech for oneself [1], the little voice in the head [2], an internalized verbal thought that can be consciously explored [3], "the subjective experience of language in the absence of overt and audible articulation" [4] or, more generally, as a form of mental imagery [5]. The unique cognitive status of inner speech is reflected in its pervasive role in a variety of functions, such as language, working memory, complex reasoning and self-awareness, among others [6-10]. For example, impairment of inner speech has been associated with the impairment of global self-awareness, self-related memories and emotional awareness, along with impaired "sense of individuality" and corporeal awareness [9].

Researchers have long recognized a methodological challenge when trying to directly and objectively investigate the highly subjective and elusive psychological processes that constitute inner speech [1,11]. An often studied proxy of inner speech in contemporary research is word phonological representation, i.e. a construct that is usually tested as a

person's ability to silently judge whether two words rhyme, whether one word is longer than the other, or whether two words are homophones [12,13]. To make silent judgments in such tasks requires the use of inner speech.

While inner speech abilities of persons suffering from post-stroke aphasia have been studied relatively extensively [12-16] there is currently no clear idea on how slow-growing lesions, such as low-grade gliomas (LGG), affect inner speech. Inner speech in post-stroke aphasia may be affected partially or completely [12-15], and typically to a degree comparable to the overt speech impairment, although cases of aphasia with discrepant overt-covert speech abilities were also reported [14]. Since the neural substrate for language responds differently to different types of lesions [17], slowly progressing brain lesions may affect inner speech abilities differently than sudden lesions such as stroke.

Diffuse LGG is a relatively rare neurological tumor [18,19]. Although it is not considered aggressive, the 5-year survival is only about 50% [20]. This type of brain tumor often invades the "eloquent" brain areas, i.e. functional areas whose damage leads to major neurological deficits e.g. Broca's and Wernicke's areas, the insular cortex, supplementary motor area, or the premotor cortex [21-23]. It infiltrates the main white matter tracts in one hemisphere and spreads

to the opposite hemisphere via the corpus callosum [18,19]. However, despite invading the “eloquent” brain areas, severe neurological deficits due to LGG are generally not observed initially [21-24]. Researchers have explained this observation in terms of the brain capacity for functional reorganization related to the affected area(s).

In the present study, we investigated whether inner speech would be affected by LGG in the left temporal lobe, which according to current theories supports inner speech abilities [25,26]. This question is clinically and theoretically important. First, continuous brain reorganization may provide sufficient functional compensation of inner speech on its own. Additionally, the effects of complex interactions between the continuous reorganization processes triggered by the tumor growth in the areas spared by the surgery and the functional reorganization of the resected area are not well understood and it is therefore difficult to make specific predictions on functional compensation in such cases. Thus, extending current knowledge on the effects of slow-growing lesions on cognitive abilities, including inner speech, is important, because it may reveal new patterns of dysfunction and functional plasticity in these patients.

Material and methods

Participants

Case presentation: We present the case of a 41-year-old man (henceforth MN, fake initials) without previous medical history, who was first seen by a neurologist in 2012 due to an isolated generalized seizure. He showed no other symptoms at that time and the general neurological examination was normal. The MRI revealed an extensive lesion in the left temporal lobe, suggestive of low grade glioma. EEG was normal. Treatment with levetiracetam 500mg/12 hours was initiated, and surgery with partial left temporal lobectomy and partial resection of the tumor was performed [27] (Figure 1).

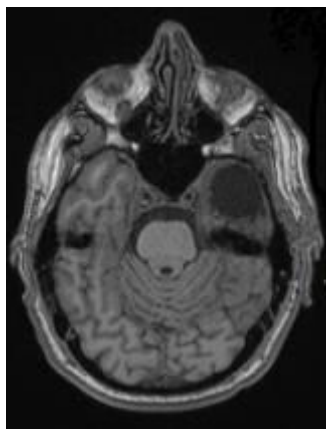


Figure 1: Axial T1-weighted contrast MRI image demonstrating lesion of the left temporal lobe. Image orientation: the left side of the brain corresponds to the right side of the image.

Anatomo-pathological analysis revealed diffuse oligodendroglioma grade II. After the surgery the patient presented with epilepsy, which was treated with three antiepileptic drugs (levetiracetam 1500mg/12h, valproic acid 500mg/8h and lacosamide 150mg/12h), and sensory aphasia. Ten months later the cerebral MRI showed signs suggestive of

progression of the tumor. An amplification of the previous left temporal lobectomy was performed without complications. Anatomopathological analysis showed areas of progression to oligodendroglioma grade III without deletion of 1p/19q. The patient also received localized radiotherapy (Figure 1). Eight months later, a follow-up MRI suggested further tumor progression, at which point treatment with temozolomide was initiated; however, radiologic follow-ups show continued signs of tumor progression, affecting the left temporal lobe and the insular white matter. The patient is currently receiving treatment with bevacizumab.

This right-handed man is a bilingual speaker of Spanish and Basque, with 17 years of formal schooling and 15 years of work experience prior to illness. Immediately post-surgery, he experienced deficits in language and memory. Most of his post-surgery disturbances with language resolved by the time of testing for the present study, except for syntactic comprehension and production, and persistent anomia. The working memory difficulties remained, despite his participation in an intensive cognitive rehabilitation program. At the time of testing, his vision and hearing were normal.

Control data: Data were collected from ten neurologically intact persons within a study by our group investigating inner speech in post-stroke aphasia (mean age 62.4 (\pm 8.8), mean of years of formal education: 12.6 (\pm 4.1)) [16]. Their cognitive status assessed by the Montreal Cognitive Assessment [28] was normal (mean 24.9 (\pm 2.2)).

Evaluative measures

As a test of general cognitive status, we administered the Montreal Cognitive Assessment [28] and we tested MN's language abilities using the Spanish version [29] of the Boston Diagnostic Aphasia Examination (BDAE) [30] and the Boston naming test [31]. We also administered verbal fluency tests (categories: clothes, tools, furniture, and fruits; letters: F, A, S, M, and N), a phonological discrimination test [32], the Month ordering test [33,34] to assess MN's verbal working memory, Raven's Progressive Color Matrices [35] to assess his nonverbal intelligence, Beck depression inventory [36] to exclude severe depression, the Edinburgh handedness inventory [37] to confirm the right hand dominance, and the Varieties of Inner Speech Questionnaire [38] to obtain insight into the patient's own awareness of his use of different forms of inner speech (e.g., dialogic vs. condense).

Experimental measures

The study consisted of four experiments assessing inner speech: silent rhyming judgments (experiment 1), syllable discrimination (experiment 2), identification of words in compounds (experiment 3) and in names for numbers (experiment 4). In addition, we tested participants' overt rhyming abilities, to exclude the possibility that a more general inability to judge rhyming would preclude successful performance on the silent rhyming test.

The silent rhyming test in our study required making silent judgments on whether a pair of words associated with a pair of drawings rhymed. This rhyming paradigm is more appropriate for the Spanish language than the classical paradigm, which requires making judgments on written words. The reason is that judging whether Spanish written words rhyme can be done visually, on the basis of words' orthography and without evoking inner speech (e.g., avión-camión). The silent rhyming paradigm with drawings has been successfully used in previous studies on inner speech in other

languages [15]. In our experiment, there were 40 pairs of drawings, of which 20 pairs represented objects whose names rhyme in Spanish and 20 pairs represented objects whose names do not rhyme in Spanish. The drawings were selected from a widely used set of stimuli [39], with the selection being based on a standardization for Spanish [40], considering name agreement, image agreement, familiarity, and visual complexity. The drawings included in the present study were chosen from the drawings with the highest ratings in Spanish speakers. The same pairs of words used in this experiment were also used to test participants' overt rhyming abilities.

The syllable discrimination task required silently identifying syllables in auditorily presented words (n=40). The stimuli consisted of sets of randomized 2-syllable (n=13), 3-syllable (n=15), and 4-syllable (n=12) highly frequent Spanish words. The third experiment required silently identifying words in compounds. The stimuli consisted of compounds (n=20) and simple words (n=20). As in the previous experiments, the stimuli included only highly frequent Spanish words and they were presented in a randomized order. The fourth experiment required silently identifying words in names for numbers. The stimuli (n=20) included trials that allowed 1, 2, 3, or 4 words to be discerned. For example, "eight" contains only one such word, whereas in "fifty-six" two additional words can be discerned: "fifty" and "six". On the other hand, Spanish doce "twelve" contains only one word for numbers, even though it is represented by two numbers, i.e. 1 and 2.

Procedures

Except for the silent rhyming test, the stimuli in the experiments were presented auditorily. The participants' responses were recorded manually on a separate response form for each experiment and scored for accuracy after the testing was completed. Self-corrections, if accurate, were counted as correct answers, but cases with more than one self-correction for the same stimulus were counted as incorrect answers. The subject was given detailed instructions on how to complete each experimental task. Crucially, he was instructed to use only inner speech and to refrain from using any strategies that would interfere with the task, such as the use of fingers to count, tapping or visual imagery. There were two practice trials before each experiment. The testing was carried out in four sessions within two weeks, in a quiet room at a local rehabilitation center. The total testing time was about 3.5 hours. The healthy control participants were tested individually, in their homes. Before testing, all participants signed informed consent. The study protocol was approved by the Basque Ethics Committee for Clinical Research and by the ethics committee of the University of the Basque Country. The study was conducted in accordance with the Helsinki Declaration guidelines on studies involving human subjects.

Results

Evaluative measures: MN achieved high scores on most evaluative tests, although the subtests involving naming, word retrieval, sentence repetition, delayed recall, and verbal working memory revealed some deficiencies (Table 1). The BDAE aphasia severity rating scale showed that his speech/language disturbances were relatively mild (his severity score was 4). The Boston naming test revealed word finding difficulty: the patient's score without prompting was 13/60, but with prompting it reached 41/60. Furthermore, MN's high score on Raven's Progressive Color Matrices (35/36) suggested intact nonverbal intelligence and his MoCA score of 22/30 suggested that the illness affected his sentence repetition ability, letter fluency, abstraction processes and delayed

recall (Table 1). He achieved 92.5% accuracy on the phonological discrimination test. Additional tests of verbal fluency confirmed the inability to retrieve more than 5 or 6 words using either letter or category cues, whereas at least 11 words per minute need to be retrieved for earning 1 point on this subtest in MoCA. Similarly, MN achieved only 25% accuracy on the verbal working memory test. Finally, the Varieties of Inner Speech Questionnaire [38] revealed that MN preferred condensed to dialogic inner speech and that he engaged more often in the form of inner speech that involves others than in motivational inner speech.

Language Evaluation Test	Score (correct/total)
BDAE: Auditory comprehension	
Word discrimination	34/36
Orientation left-right	8/8
Body parts	13/18
Sequences of orders	5/5
Ideational material	19/24
BDAE: Speech production	
Oral agility non-verbal	6/6
Oral agility verbal	7/7
Automatized sequences	35/36
Word repetition	10/10
Repetition of phrases	15/16
Word reading	10/10
Answering questions	10/10
Naming to visual confrontation	26/32
Body parts	5/10
Verbal fluency: animals	9 (1.5 min)
BDAE: Reading and writing	
Reading aloud	10/10
Discrimination of letters and words	10/10
Written word recognition	8/8
Oral spelling	8/8
Reading sentences and phrases	10/10
Word-picture matching	10/10
Writing	5/5
Writing to dictation	15/15
Boston naming test	
Correct without cueing	13/60
Correct with semantic cuing	2/29
Correct with phonemic cueing	27/29

Total correct with cueing	29
Total correct	41/60
Additional Tests	
Raven's Progressive Color Matrices	35/36
MoCA - total	22/30
MoCA – sentence repetition	0/2
MoCA – letter fluency ("P")	0/1
MoCA – abstraction	1/2
MoCA – delayed recall	1/5
Semantic working memory	5/20
Phonemic discrimination	37/40
Beck depression inventory	2/40
Additional verbal fluency tests – 1 min	
Category: clothes	5
Category: tools	0
Category: furniture	5
Category: fruits	5
Letter: F	5
Letter: A	5
Letter: S	5
Letter: M	9
Letter: N	4
Varieties of inner speech questionnaire	
Dialogic	9 (4-24)
Condensed	21 (5-30)
Other people in inner speech	22 (5-30)
Evaluative/ motivational	10 (4-24)
Experimental Tests	
Rhyming - silent	27/40
Rhyming – overt	40/40
Syllable discrimination	40/40
Words in compounds	38/40
Words in numbers names	18/20
BDAE- Boston Diagnostic Aphasia Examination; MOCA– Montreal Cognitive Assessment.	

Table 1: MN's performance on evaluative and experimental tests.

Experimental measures: The modified t-test, which was designed for comparing single cases to small samples [41], revealed that MN's silent rhyming was considerably worse ($p=0.027$) relative to the HC group,

whereas his performance on the remaining tests was comparable to the performance of HCs (overt rhyming: $p=0.136$; words in compounds: $p=0.288$, discerning syllables: $p=0.268$; discerning words in names for numbers: $p=0.48$). All tests were one-tailed, as recommended [41].

Discussion

The most striking finding of the present study is that inner speech appears to be relatively preserved in this patient, despite the slowly-progressing tumor in the left temporal lobe. The relatively preserved inner speech abilities are indicated by the patient's high scores on all inner speech tests, except for the silent rhyming test. Given his 100% correct performance on the overt rhyming test, his performance on the silent rhyming test indicates that MN does not have a difficulty with rhyming itself. Instead, his difficulty with silent rhyming is likely due to the word retrieval deficit. Namely, the task in our study required judgments on whether two words associated with a pair of drawings rhymed (section "Experimental measures"). Thus, the task required retrieval of words referring to the depicted objects. We speculate that the difficulty was related either to lexical selection, i.e., the problem of choosing the correct two words that would match the presented pair of drawings from a set of retrieved words, or to the deficit in word retrieval more generally, i.e. the problem in retrieving any words, which would preclude the possibility to make judgments on whether the words in fact rhymed or not. The interpretation that MN's silent rhyming was affected by a word retrieval deficit is compatible with his poor performance on the Boston naming test and verbal fluency tests (section "Results"). Taken together, the data indicates that MN has impaired word retrieval, with the deficit likely affecting both lexical selection and phonological word form. This conclusion is compatible with the fact that MN's lesion is in the left temporal lobe, affecting Wernicke's area, which supports phonological word form, and the neighboring areas (mid to posterior middle temporal gyrus), which support lexical access [25].

An alternative explanation of MN's poor performance on the silent rhyming test hinges on the concept of limited verbal working memory capacity. The working memory load in the silent rhyming task is relatively high, because the task requires interpreting pairs of drawings, keeping track of the semantic information derived from the drawings, i.e. the concepts associated with the objects depicted in drawings, retrieving appropriate words, and maintaining the retrieved words in working memory while judging whether they rhymed. MN's low scores on the subtests of MoCA related to language and memory for language (e.g., repetition of sentences, delayed recall of words, and verbal fluency) as well as his poor score on the test of verbal working memory suggest that this explanation is also plausible. However, the limited verbal working memory capacity does not fully explain MN's performance on the silent rhyming test, even though it might have exacerbated the difficulty with word retrieval.

An intriguing question arising from the present study pertains to the possibility of engaging in various forms of inner speech when word retrieval is compromised. Current models postulate that inner speech repairs (i.e. internal self-monitoring) take place at a later stage in speech production, i.e. after retrieval of the phonological word form [25]. These models predict that, unlike the speech errors that occur within a later time window [42] and can be self-monitored internally, errors in lexical selection cannot be remedied in inner speech. The effects of such errors on cognition, self-awareness and consciousness deserve more attention, given the pervasive role of inner speech in human mental life.

In conclusion, MN's inner speech appears to be affected by the word retrieval deficit, like his overt speech. The present study's finding that inner speech appears largely preserved in this case of slow-growing tumor in the left temporal lobe is consistent with other findings on the brain's remarkable potential for plasticity [43]. In grade II gliomas, the rate of continuous tumor growth is slow, amounting to 5 mm per year [44]. The slow growth of the lesion allows a continuous, slow reorganization of the functions at risk for impairment. The brain accomplishes functional reorganization by engaging the neighboring ipsilateral and/or homologous contralateral areas. As an example, a substantial resection of grade II glioma in the left inferior frontal gyrus, which is one of the classical language areas, did not result in severe language deficits [45], as would be expected if the functional localization of the brain were fixed. Similar findings were reported after removal of the whole left dominant insula [46] and Wernicke's area [47]. Our data further support this insight, by providing evidence on relatively spared inner speech in a patient with partially removed left temporal lobe tumour, which had progressed beyond stage II at the time of testing.

To our knowledge, this is the first study that explicitly investigated the impact of left temporal slow-progressing tumor on inner speech. Future studies involving large samples and using neuroimaging will identify changes in patterns of activation associated with inner speech in patients with slow-growing lesions in the left temporal lobe. Finally, studying inner speech in different neurological profiles is important, not only because it contributes to a better understanding of inner speech deficits, but also because it opens window into a unique cognitive ability that is implicated in various aspects of human mental life. Given the critical role of inner speech in language, memory, reasoning, emotional awareness, self-awareness and other cognitive functions, assessment of this ability in patients harboring tumors in the left temporal lobe is highly recommended in clinical settings.

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Conflict of Interest

The authors declare that they have no conflict of interest.

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