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Autobiographical memory and the self in a single-case of chronic unilateral spatial neglect

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ABSTRACT

Unilateral spatial neglect (USN) is mainly defined as a condition affecting perception and the mental representation of the environment. However, nothing is known about its impact on the ability to mentally represent one's past and on personal identity. We addressed these questions in a case of chronic USN, DR, a 59-year-old right-handed woman, who underwent a variety of measures exploring the self and autobiographical memory (AM). DR showed preserved self-images and her AM performance was only preserved when memories were prompted by her own self-images and not by self-unrelated cues. Our findings are discussed in light of the interconnection between the self and AM.

KEYWORDS : Autobiographical memory; self; unilateral spatial neglect; cognitive neuropsychology; single-case study

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Introduction

The ability to remember one's past (i.e., autobiographical memory; AM) is of fundamental importance for the experience of enduring as an individual over time but also for social relationships and for guiding behaviors (Conway, 2005). Previous research in cognitive neuropsychology highlights the relevance of exploring AM in clinical conditions because of its impact on daily life functioning, for instance leading to distress in social situations or frustration when not being able to remember (Ernst et al., 2014; Rathbone, Moulin, & Conway, 2009).

According to the Self-Memory System model (SMS; Conway, 2005), AM and the self share a bidirectional relationship. As such, the self has an organizing role in memory and in return, identity formation is supported by a set of self-defining memories. Within this theoretical framework, Rathbone, Moulin, and Conway (2008) developed the I AM task, which operationalized the self as "self-images" (i.e., enduring and important facets of the self, including physical, psychological, or social aspects). Using this task, clinical findings support that the study of the self is critical in understanding how people cope with their condition and how this impacts on their psychological well-being (Rathbone, Holmes, Murphy, & Ellis, 2015). However, while a loss of or a reduction in the accessibility of self-images has been described in clinical populations as a result of AM impairment (e.g., Addis & Tippett, 2004; Tanweer, Rathbone, & Souchay, 2010), other studies suggest that in some cases, self-images can be maintained by semantic components of AM, despite deficient episodic AM (Illman, Rathbone, Kemp, & Moulin, 2011; Rathbone et al., 2009).

We report here the case of a patient with chronic left unilateral spatial neglect (USN) - a syndrome that commonly arises after lesions of the right hemisphere and which is characterized by disturbances in the ability to detect, identify and move toward objects or even to have a mental representation of the contralesional (left) side of space (Saj, Fuhrman, Vuilleumier, & Boroditsky, 2013). Most patients show a rapid recovery of USN but, in rare cases, this syndrome persists and may be regarded as chronic (Stone, Patel, Greenwood, & Halligan, 1992).

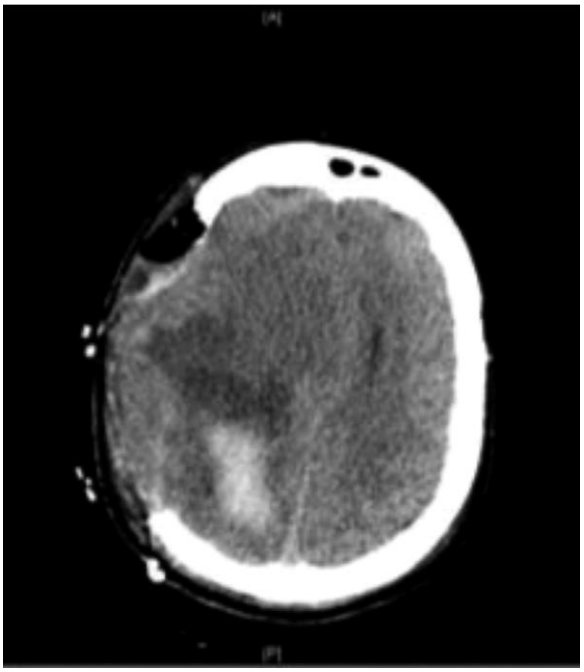
While studies on USN generally focus on perception and attention, which are at the forefront of this

condition, one could reasonably expect that USN might impinge on AM. Indeed, since USN compromises both the perception and the mental representation of the environment, it is likely that the mental representations of the past will also be affected, especially since personal memories are characterized by their visual content (Conway, 2005). In this vein, Saj et al. (2013) explored whether people with left USN would have difficulty representing events that fall to the left on the spatial representation of the mental time line, a hypothesis which was supported by their findings. However, this study did not focus on patients' personal memories per se but on the recall of past and future characteristics from a fictitious person. This led us to explore AM for the first time in USN, and specifically, how personal past events and the self are mentally represented in a single-case with chronic USN.

Case report

DR is a 59-year-old right-handed woman, who was working as an architect until she suffered from a hemorrhagic stroke in December 2013. She was under the care of a neurologist at the University Hospital of Dijon (France) and neuroimaging showed a right frontoparietal lesion (Figure 1).

Figure 1. DR's CT scan 15 days after her stroke (January 2014) showing right frontoparietal lesion traces (the right is on the left).



One month after her stroke, DR underwent a comprehensive neuropsychological assessment. Scores obtained by DR compared to normative data for each test are summarized in Table 1. This neuropsychological assessment revealed a left USN, a left hemianesthesia and attentional disorders (i.e., alertness, as measured by the alertness subtest from the Test of Attentional Performance (TAP 2.2). Her USN was mainly egocentric, with problems in the ability to detect and to take into consideration objects on the left side of space in the visual, auditory, and somesthetic modalities. At this stage, the neuropsychological examination also revealed mild impairment in executive function, specifically in verbal fluency (i.e., a measure of executive control ability), and DR also had borderline score for inhibition. Anterograde memory scores were also at the limit of the normal range but language and praxis were preserved. While DR had no particular subjective complaints, her husband had noticed occasional forgetting in daily life. After 24 weeks, only her attentional impairment and USN persisted, despite a slight improvement. Her USN was thus considered as chronic (note that her last follow-up examination conducted in June 2015 - i.e., after the testing in this study was carried out - confirmed that her USN was still present). In comparison, there is a relative preservation of her general cognitive functioning. The AM assessment described below was carried out 15 weeks after DR's stroke.

Autobiographical memory: test episodique de la *mémoire* du *passé autobiographique (TEMPau)*

To assess AM, DR underwent the TEMPau (Piolino, Desgranges, & Eustache, 2000), which consists of generating a total of 24 specific memories across five lifetime periods (0-17 years old, 18-30 years old, more than 30 years old except the last 5 years, last 5 years except the last 12 months, and the last

12 months) and providing as many details as possible about the memories. For each life period (except the last year), participants have to retrieve memories related to four generic cues (e.g., relationships, school, work, travel). For the last year period, eight generic cues are used (e.g., last summer, last week, etc.) using a chronological approach. While no practice session was conducted, examples of specific and non-specific memories were provided to ensure that DR understood this distinction. No time limit was set.

Each memory is rated on a five-point scale (0 = absence of memory or general information to 4 = specific and richly detailed memory). DR's scores were compared to the French normative data for this test (Piolino, 2008) for the overall AM score. This included all the scores whatever their nature (max. 4x4 = 16) and also the strictly episodic score (i.e., including only the specific detailed memories, that is memories rated as 4 (max. 4x4=16).

The TEMPau performance is summarized in Table 2. Crawford and Howell's method (1998) was applied for all the statistical comparisons. Compared to norms, DR obtained a lower overall AM mean score and a lower strictly episodic mean score. For the overall AM score, this impairment seemed present for all the life periods, except for the last five years. For the strictly episodic score, of the 24 memories requested, DR only provided two episodic memories; the remaining were semantic in nature and her performance seemed particularly low for the 18-30 and the last five years' periods. Examples of memories provided by DR during the TEMPau are available in Supplemental Data.

I AM task

To explore the relationship between AM and the self, we carried out the I AM task (Rathbone et al., 2008). In the absence of normative data for this test, DR performance was compared to those of 14 healthy controls (10 women) matched for age (mean: 65.71; *SD:* 19.55). This test consists first in generating as many self-images as possible that are important in defining current identity (e.g., *I am a mother, I am shy).* Identity statements were scored using the modified coding scale from the twenty statements test (TST; Rhee, Uleman, Lee, & Roman, 1995). Each statement was coded according to eight categories (e.g., traits, social identities, physical descriptions, etc.) and this allowed us to measure the complexity of identity (i.e., the number of categories sampled in participants' responses). Participants have to select the three most important or significant of their self-images and to provide for each one, three specific memories and to describe them in as many details as possible. To ensure understanding of instructions, examples of self-images and associated memories were provided to all participants.

A similar procedure for the TEMPau was used to score these memories (i.e., a five-point scale going from 0 = absence of memory or general information to 4 = specific and richly detailed memory). A second rater scored 34% of the memories, showing strong inter-rater reliability (Kendall's correlation coefficient: 0.85).

DR and healthy participants' scores are summarized in Table 2. Using the Crawford and Howell's method (1998), we observed that DR provided a similar number of self-images as healthy participants, suggesting a preserved ability to define herself (examples of DR's self-images: *I am a woman, I am hard-working, I am reserved).* Qualitatively, all the self-images and associated memories provided by DR applied to her before her stroke. In addition, no significant difference was found between DR and the control group regarding the complexity of identity or the overall AM score of memories triggered by the participants" own self-images.

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Table 1. DR's scores at the neuropsychological baseline examination.

	DR's score	Normative data[a]
USN		
Line bisection (BIT; mean deviation from the midpoint, in mm)	-3	<±6.5 mm
Target/distractors research (BIT)		
• Star cancellation (number of cancelled stars)	20/54 [b] (left-side neglected)	>44
• Star ratio	0.00[b]	>0.46
• Letter cancellation (E/R; number of cancelled letter)	9/40[b] (left-side neglected)	>32
Ogden figure (raw score)	3/4[b] (left-side neglected)	≤0
ROCF (raw score)	14/36[b] Left-side neglected	Z = - 4.8
Catherine Bergego scale (raw score)	17/30[b]	Scores between 11 and 20: moderate USN
Visual extinction (BEN; number of extinctions)	Left extinction (three extinctions on four bilateral stimulations)[b]	Any extinction
Neglect subtest (TAP 2.2)	Left:8/24[b] (1494 ms)	Pc. < 1
• Scores (mean RT)	Right: 19/24[c] (597 ms)	Pc. 8
Text reading (presence of start of lines omission)	Start of line omission (left-side neglected)[b]	Any omission at the left
Anterograde memory		
Hopkins verbal learning test, form 1 (number of words)		
• First recall	5/12[c]	Pc. 5
• Second recall	7/12[c]	Pc. [5-16]
• Third recall	8/12[c]	Pc. 5
• Differed recall	7/12[c]	Pc. [5-16]
• Total recall	20/36[c]	Pc. 5
• Recognition	12/12	Pc. > 50
Executive functions and attention		
Digit span (WAIS-IV; scaled scores)		
• Forward	6	Scaled score: 10
• Backward	5	Scaled score: 9
Verbal fluency (GREFEX; number of words)		
• Letter	17[c]	> 15
• Categorical	20[b]	> 24
GonoGo (TAP 2.2)		
• Mean total RT	517 ms[c]	Pc. 12
• Errors (number)	0	Pc. > 62
Alertness (TAP 2.2)		
• Mean RT without alert	605 ms[b]	Pc. < 1
• Mean RT with alert	840 ms[b]	Pc. < 1
• Phasic alert index	-0.35[b]	Pc. 1
Language		
Bachy-Langedock 36 (raw score)	36/36	>35
Praxis		
Mahieux's battery (raw score)		
• Expressive gestures	5/5	>4
• Use of objects mime	10/10	>8
• Meaningless gestures	8/8	>7

RT: reaction time; BEN: Batterie d'Evaluation de la Négligence; BIT: behavioral inattention test; MEM-III: Wechsler Memory Scale-III; ROCF: Rey-Osterrieth complex
figure; *SD:* standard deviation; TAP 2.2: Test of Attentional Performance 2.2 version.
[a]The normative data are based on the established norms for each test, corresponding to DR's age. Depending on the test, the normal range is expressed as a z-score (pathological z-score = -1.65), a percentile (pathological percentile: Pc < 5), or a cut off score.
[b]Score under the normal range,
[c]borderline score but remaining in the normal range.

Discussion

This single-case study explored AM and the self in chronic USN for the first time (to the best of our knowledge). While perceptual and attentional abilities are generally the critical domains in this condition, our data suggest that an AM deficit could be present in chronic USN, in the context of a relative preservation of other cognitive functions, such as language or anterograde memory. Despite AM impairment, DR provided without difficulty a series of self-images that define herself, with similar qualitative properties as healthy participants. While AM and the self-share a strong relationship at the theoretical level (the SMS model; Conway, 2005), our results fit with the growing body of clinical research using the I AM task showing that AM impairment does not consistently lead to a loss or an impoverishment of the self. For instance, previous studies have documented that the self can be supported by semantic aspects of AM when episodic AM is deficient (Illman et al., 2011; Rathbone et al., 2009). However, DR's profile of AM results advocated an alternative view, since the memories supporting her self-images were not semantic or overgeneralized.

In particular, an intriguing finding is the different benefit from memory cueing reported in DR, who showed preserved AM performance when memories were cued by her own self-images but not when memories were prompted by generic/self-unrelated cues. In other words, DR provided memories with a similar episodic quality as those from healthy participants when triggered by her self-images. Based on the SMS model, we suggest cautiously that DR's results could be related to retrieval mechanisms. Specifically, the use of self-images as cues to elicit memories could be less demanding in terms of retrieval strategies, with thus an easier access to memories, than with the use of generic cues. In support, such a finding could be related to the self-reference effect, which corresponds to the tendency for people to better remember information relevant to them (see Klein, 2012 for a review). Recently, Kwan, Kurczek, and Rosenbaum (2015) reported similar outcomes in amnesic patients when prompted by personal cues derived from previous past events. Kwan and colleagues suggested that the benefit from personal cues could be related to the activation of some kind of framework conveyed by self-relevant cues, which could then help to structure event details. Regarding the current study, and in relation with the SMS model (Conway, 2005), the activation of self-images in the working self (i.e., a dynamic system containing goal structures that control, among others, the retrieval of specific memories) would have an organizational function in AM. In particular, self-images would activate, through the working self, a set of highly accessible memories supporting each self-image (Rathbone et al., 2008). In contrast, the use of generic cues would require more effortful processes with a first elaboration of the cue, then a further search in memory, then the selection and assessment of the retrieved memory.

From a neuroanatomical standpoint, DR suffered from frontoparietal damage, a brain region involved in AM retrieval (Berryhill, Phuong, Picasso, Cabeza, & Olson, 2007; Svoboda, McKinnon, & Levine, 2006). In addition, according to the SMS model, the frontal lobe is particularly involved in effortful retrieval, which fits well with DR's impairment when memories were prompted by generic cues that require more retrieval strategies.

Needless to say, alternative hypotheses could also account for DR's AM impairment. This includes the presence of attentional disorders (i.e., alertness in this case) or the role of the inferior parietal lobule in conscious awareness (Cabeza, Ciaramelli, Olson, & Moscovitch, 2008). However, these two alternative suggestions fit less with the differential effect of self-related vs. generic cues on DR's performance and further studies are necessary to support our current hypothesis.

In a previous study, Saj and colleagues (2013) showed that patients with USN have difficulties representing past events along a mental time line. As stated earlier, their study did not focus on patients' personal memories. While our results support to some extent the impact of USN on the ability to form mental representations of the past, an open question is whether Saj et al.'s results would be different if people with USN have to represent past events along an individualized mental time line, built on personally meaningful temporal labels or milestones (e.g., my wedding, the birth of my child, my hospitalization). Also of interest, Saj and colleagues showed that the difficulties in representing events along a mental time line are restricted to the past and do not extend to the representation of the future. Considering that AM and episodic future thought (i.e., the ability to imagine personal future events) share a strong relationship - notwithstanding that episodic future thought requires additional cognitive and neural resources (Schacter et al., 2012 for a review), a future direction would be to explore if our current findings for the past would be similar for the future.

From a clinical standpoint, this single-case provides new insights into the cognitive profile of people with chronic left USN. Needless to say, further studies are warranted to explore whether this AM

profile is observed in other cases of USN (in both transitory and chronic cases) and to disentangle the precise relationship between the characteristics of USN and this profile of AM impairment. In particular, a critical issue would be to compare memories with matched temporal distance that were encoded before or after the occurrence of USN - an issue that we could not discuss in DR's case due to the short time frame between her stroke and AM assessment. A limitation of the current study that should also be addressed in further studies would be to have patients' self-images checked over by their relatives.

More generally, these findings support the usefulness of AM assessment in clinical practice to reveal memory impairment undetected by routine anterograde memory tests, but which could have a deleterious impact in daily functioning of patients. These results also reflect the occurrence of subtle AM impairment in patients, whose observation might depend on the method of assessment used and, more specifically, on the cue-words used to elicit memories. Since the cue-word method is one of the most frequently used methods of AM assessment, this suggestion could have implications for future clinical research and the development of sensitive diagnostic approach. In addition, we echo the recent recommendations by Kwan et al. (2015), who promoted the overall contribution of these kind of studies in creating more effective ways to structure questions in a personally meaningful way, to interact with people with memory impairment. Whether these findings might be a basis for the development of cognitive interventions aiming to support AM represents an avenue for future research.

Table 2. Scores obtained by DR and healthy participants at the TEMPau and the I AM task.

	DR	Healthy participants mean *(SD)*	Statistical analysis[a]
TEMPau [b]			
Overall AM score			
° 0-17 years	7 *	13.18 (2.41)	t = -2.52, p = 0.02
° 18-30 years	9**	14.00 (1.87)	t = -2.63, p = 0.01
° more than 30 years old	8*	13.63 (2.38)	t = -2.33, p = 0.02
° the last 5 years	9	13.35 (2.38)	t = -1.80, p = 0.08
° the last year	9**	14.21 (1.73)	t = -2.96, p = 0.005
Total (mean score)	8.4*	13.67 (0.43)	t = -12.07, p < 0.001
Strictly episodic AM score			
° 0-17 years	0	8.71 (5.25)	t = -1.63, p = 0.11
° 18-30 years	0*	10.35 (4.19)	t = -2.45, p = 0.02
° more than 30 years old	4	9.41 (4.39)	t = -1.21, p = 0.23
° the last five years	0*	9.41 (4.39)	t = -2.11, p = 0.04
° the last year	4	10.59 (4.28)	t = -1.51, p = 0.13
Total (mean score)	1.6*	9.69 (0.77)	t=-10.35, p < 0.001
I AM task			
Number of self-images	10	8.14 (3.68)	t = 0.49, p = 0.63
Complexity of identity (number of categories used)	3	3	χ^2 = 0, p = 1
Overall AM score	18	19.43 (5.98)	t = -0.23, p = 0.83

[a] t-Tests based on the Crawford and Howell's method (1998) for single-case studies.
[b] The healthy participants' scores mentioned in this table for the TEMPau correspond to those of the normative database from Piolino (2008; n = 34).
* Significant difference at p = 0.05; ** Significant difference with an adjusted p-value (Bonferroni correction: $\alpha/5$) of p = 0.01 for the t-tests ran on the five life periods.

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Disclosure statement

No potential conflict of interest was reported by the authors.

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