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Illustrating where spatial perception versus memory-based representation: spatial neglect in a distinguished artist; a case report

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Abstract

Spatial neglect is a disorder of attention, perception, and processing of stimuli in contralesional space. The heterogeneous behaviors involve diverse neuroanatomical mechanisms. Anecdotal evidence suggests that neural circuitry of active spatial perception may differ from that used to encode spatial memory. These differences can escape detection by conventional assessments, thereby leading to missed diagnoses. We present a case, in an artist who demonstrates selective impairment in a contralesional space during active Where spatial perception. His performance is better when asked to draw entirely from memory. This case highlights the variability in neglect, importance for comprehensive testing, and encourages further investigation.

Keywords

Cerebrovascular accident; spatial neglect; artists; representational neglect

Introduction

Approximately 798,000 people in the United States have a stroke each year (Mozaffarian et al., 2015). Of these, 350,000 (30–70% of right-brain stroke survivors and 20% of left-brain stroke survivors) present with a cognitive disorder, known as spatial neglect (Bowen & Lincoln, 2007; Katz, Hartman-Maeir, Ring, & Soroker, 1999). Although the presentation of spatial neglect differs widely, it is defined as the inability to attend to, perceive, process, or act on stimuli in contralesional space, which results in functional disability, poorer rehabilitation outcomes, and an overall longer recovery (Chen, Hreha, Kong, & Barrett,

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2015; Heilman, Valenstein, & Watson, 2000; Nijboer, van de Port, Schepers, Post, & Visser-Meily, 2013).

Historically, spatial neglect has been underdiagnosed, possibly because of its diverse behavioral presentation (Bowen, McKenna, & Tallis, 1999). It was reported that using more than one test may decrease the false negative rate and optimize delivery of care (Azouvi et al., 2002; Azouvi, 2017). This may be because spatial neglect may present differently in different individuals and conventional assessments may be limited to only certain areas of spatial performance (Bartolomeo, 2007). For example, the Apples test is widely accepted to assess for allocentric (object-centered) spatial bias (Ota, Fujii, Suzuki, Fukatsu, & Yamadori, 2001). There is an argument that the National Institutes of Health (NIH) Stroke Scale may be sub-par when assessing for spatial bias (Hillis, Wityk, Barker, Ulatowski, & Jacobs, 2003). Additionally, our group and others have previously reported on assessments, including the Behavioral Inattention Test (BIT), to be better in detecting Where (perceptual-attentional) spatial bias, as opposed to Aiming (motor-intentional) spatial bias (Goedert et al., 2012; Na et al., 1998).

Despite these repeated demonstrations, most clinicians only use quick assessments (e.g., clock drawing) for the detection of spatial neglect. Research studies have however included detailed neuropsychologic assessments to identify neglect behaviors (Cicerone et al., 2000; Paolucci, Bureca, Multari, Nocentini, & Matano, 2010). Additionally, even for those diagnosed in the acute phase, different types of spatial bias may respond differently to interventions (Goedert, Chen, Boston, Foundas, & Barrett, 2014). Overall, this work emphasizes the need to accurately diagnose and classify patient's symptoms prior to starting therapeutic treatments.

Specifically, it may be very important to distinguish between active Where spatial perception and memory-based representation to better enhance rehabilitation outcomes (Coslett, Bowers, Fitzpatrick, Haws, & Heilman, 1990). Where spatial perception is associated with perceiving or attending to real-time stimuli and has previously been shown to mediate directional action, response inhibition, persistence, and motor/personal self-regulation (Coslett et al., 1990; Na et al., 1998). Bias in this network can be measured by tasks that require cancellation or figure copying. In contrast, tasks that require drawing from memory may better assess neural networks used for storage/recall of spatial information.

One way to distinguish between these processes is to examine the presentation of spatial neglect in artists (Cantagallo & Della Sala, 1998; Marsh & Philwin, 1987). Artists use complex memory-based representational systems when they engage in the creation of a masterpiece. For example, a German artist, Lovis Corinth, said: "true art means to use unreality"; in essence: "true art" is that which is produced only from the artist's memory (Bazner & Hennerici, 2006). Since artists engage in this process frequently, mechanisms of memory representation may be more resistant to hypoperfusion during stroke in these individuals (Cantagallo & Della Sala, 1998; Marsh & Philwin, 1987). Here, we examine this possibility and question its implications for the diagnosis.

Case report

A.R. (not his real initials) is a 68-year-old, right-handed, Chinese-American male artist with a history of hypertension. During his internationally renowned career, A.R. painted for local venues, participated in gallery showings, achieved numerous awards of distinction, and published two books containing his artwork. He continued to be an avid lover of painting until the evening of 25 April 2012, when his wife found him with left-sided weakness, a right gaze preference, and slurred speech. He was diagnosed with a large right middle cerebral artery (MCA) stroke involving the frontal, parietal, and occipital lobes (Figure 1). There was a small hemorrhagic conversion with cerebral edema and midline shift.

At the acute hospital, he was given tissue plasminogen activator and also had a Mechanical Embolus Removal in Cerebral Ischemia (MERCI) clot retrieval procedure in order to help with MCA revascularization. His initial neuromuscular and skeletal physical examination was significant for impaired left facial droop, right gaze preference, and left-sided weakness. He was alert and oriented to person, place, and time. His medical doctor treated him with heparin and aspirin as a preventative measure for recurrent stroke. A.R. was not diagnosed with spatial neglect because of normal performance on clinical measures, such as the Function Independence Measure score and the NIH Stroke Scale, and thus, no spatial neglect therapies were recommended. However, in the clinical documentation notes there are specific wordage that directly suggests that A.R. did in fact have some form of spatial neglect. For example, his occupational therapist wrote that he had a gaze preference toward the right side and required verbal cues to turn his head and move his body toward the left. This presentation shows two very common neglect behaviors. Ipsilesional directed gaze, meaning that the person is looking away from the direction of the neglected space (or toward the lesion), can range in severity; however, it is frequently observed. This head deviation is not because of a muscle imbalance or purposeful but rather due to an attention network problem (Corbetta & Shulman, 2011; Mesulum, 1999). Additionally, the directed body movements are usually difficult to conduct in the contralesional space (neglected or space opposite to the stroke lesion) (Barrett & Muzaffar, 2014). He also demonstrated not eating food on the left side of his plate, on multiple occasions and not being aware of this error. Anosognia is also a common problem associated with spatial neglect (Azouvi et al., 1996).

Assessment of spatial neglect

Approximately 1-year post stroke, A.R. visited an outpatient occupational therapist, because he was interested in starting preventative therapies. At this time he displayed occasional asymmetric behavior when attending to or initiating movement toward leftward stimuli. The therapist then tested A.R. for neglect-related functional disability using the Catherine Bergego Scale via Kessler Foundation-Neglect Assessment Process (KF-NAP™) (Chen, Hreha, Fortis, Goedert, & Barrett, 2012). A.R.'s performance was consistent with mild neglect. Specifically, these were his results: "gaze orientation" a score of 1, "limb awareness" a score of 2, "grooming" a score of 2, "navigation" a score of 0, "dressing" a score of 0, "personal belongings" a score of 0, "auditory attention" a score of 0, and "collisions" a score of 1. There were two items that were not able to be scored: "eating" and "cleaning after meal". This was because A.R. was medically unable to consume any food or

drink through his mouth and required total parenteral nutrition. Thus in order to calculate the final score, we took the total score (which was 6), divided it by the items scored (8 items), and then multiplied that score (which was = 0.75) by 10. The final score is a 7.5 or mild neglect. The final score calculation supports the integrity of the diagnosis because the items are all one factor (Chen, Chen, Hreha, Goedert, & Barrett, 2015).

After the KF-NAP assessment was positive for spatial neglect, A.R. was referred to our stroke research team and underwent additional neglect screening with the (1) BIT and (2) five-element drawing test (Gainotti, Messerli, & Tissot, 1972; Wilson, Cockburn, & Halligan, 1987). He was also tested for visual extinction and hemianopia, both of which were negative. We found that A.R. exhibited clear neglect symptoms on tasks that required copying objects (actively based Where perceptual-attentional spatial bias), primarily the BIT figure copying section and line bisection, in addition to the five-element drawing test (see Figure 2). However, A.R. exhibited less asymmetries on tasks that required drawing from memory (memory-based representational spatial bias), for example the BIT face and butterfly drawing section (see Figure 3).

Discussion

Our participant A.R. was initially not diagnosed with spatial neglect, possibly because of a likely lack of comprehensive initial assessment at the acute hospital. Difficulty with comprehensive testing may be due to short length of stays at acute care hospitals, lack of routine clinical procedures, or because of time constraints that therapists can have. However, after reviewing the hospital reports (including clinical notes), it seemed conclusive that A.R. did have signs of a spatial neglect syndrome. One year later, when his spatial performance was formally assessed in outpatient therapy, he did in fact have spatial neglect. It was mild however affecting his functional performance. A.R. was referred to therapy, not due to an acute event but rather because his family wanted him to start therapy in order to prevent functional decline and improve his overall quality of life. Thus, no new imaging exams were taken at the 1-year post-stroke time point. Despite the lack of imaging comparing A.R.'s initial and 1-year visit, his behavioral performance 1 year later still leads us to hypothesize that neural networks underlying spatial performance may be selectively active during certain behaviors. For example, there were differences in performance when A.R. was engaged in cancellation or figure-copying tasks (Where perception) versus drawing from memory (memory representation).

This case report, categorizing deficits of spatial neglect in an artist, emphasizes the need for comprehensive neglect testing that can assess for a diverse array of these networks to reduce missed diagnoses and treatment delays. And potentially, having clinical procedures for assessment of neglect will reduce the large variability of prevalence (30–70%) for right brained stroke patients who present with spatial neglect. Further, evaluating the predominant type of network affected in an individual's stroke may better target therapies and enhance rehabilitation outcomes.

In a prominent previous publication, the researchers argued that artists, because of their frequent training and repetition, may have highly developed neuroanatomical mechanisms

for spatial perception and memory (Cantagallo et al., 1998). Our hypothesis is consistent with this argument. However, the categories used for evaluation of their patient, the internationally renowned Federico Fellini, were significantly different from the ones used here. The first category, visuo-perception, consisted of tasks such as reading and line bisection. The second category, visuo-motor, consisted of tasks such as cancellation, figure copying, and drawing. The authors found Fellini demonstrated spatial impairment only on visuo-motor tasks, and not those involving visuo-perception.

We did not test visuo-perception using reading or line bisection. Instead, we hypothesized that the tasks used in their visuo-motor category may be grouped into those requiring active Where spatial perception and those requiring memory representation. According to Cantagallo and DellaSala's raw data reports, Fellini demonstrated selective impairment only on cancellation and figure-copying tasks. His performance on the Rey's figure memory tasks was non-pathologic. In addition, the authors present many figures that the Fellini drew from memory post-stroke. Though he did omit some particulars on the left side and tended to draw on the right side of the page, Fellini's spatial bias on these tasks was rather mild.

Our findings are in line with their results; our participant A. R. also demonstrated clear signs of spatial neglect only on cancellation and figure copying, tasks that require more active Where spatial perception. His performance on tasks requiring drawing from spatial representation in memory was not pathological, even though there was some asymmetry.

The results of these two case reports can be summated to show that artists provide a unique opportunity to differentiate for spatial mechanisms. In general, these individuals, post right hemispheric stroke, are better at drawing from memory than engaging in cancellation tasks. One possible explanation for this may be that the repetitive and creative nature of an artist's profession causes enhanced neurogenesis and thus increased angiogenesis in various parts of the cortex. Thus, the increased collateral vessels may make a patient more resilient to hypoperfusion during stroke. There are currently promising studies that show enhanced neurogenesis in mice that have been cognitively challenged, particularly seen in post-stroke mice (Xiong, Mahmood, & Chopp, 2010). However, while this finding is strongly in support of this hypothesis, future research using brain imaging is indicated to confirm this theory.

Since clock drawing is the most common bedside test used by clinicians for the diagnosis of spatial neglect, it is very likely that individuals with training in artistic ventures, like A.R. and Fellini, may escape detection. This would lead to significant chronic impairments and increased healthcare costs. Thus, it is vital to routinely assess for spatial neglect and use a comprehensive battery to ensure detection of the complexity of the behaviors, even if the person does not show observable signs of neglect. Clinicians have reported that time is always a barrier. Thus, we recommend using a functional assessment like the KF-NAP (tests for egocentric neglect) and also the five-element drawing test (tests for allocentric neglect) could improve the detection rate and thus offering people more chances to have treatment opportunities.

Conclusion

Our facility is fortunate to be equipped with a dynamic stroke research center that collaborates well with our hospital team, which led to the detection of Where perceptual errors that A.R. made on neglect assessments. By sharing this case report, we emphasize the need for comprehensive testing that is able to evaluate a range of spatial perceptual networks in order to make the diagnosis of SN prompt and accurate. Specific assessments may prevent missed diagnoses for individuals with artistic talent or any other individuals who may be cognitively stimulated in these two pathways.

Further research should not only improve diagnostic testing, but also investigate into therapies that target-specific networks affected in individuals with right hemispheric stroke, thereby enhancing rehabilitation in people with spatial neglect. Studying the trends of recovery in these patients coupled with assessment of modern imaging techniques would additionally contribute to our understanding of neuronal repair and the link between neurogenesis and angiogenesis.

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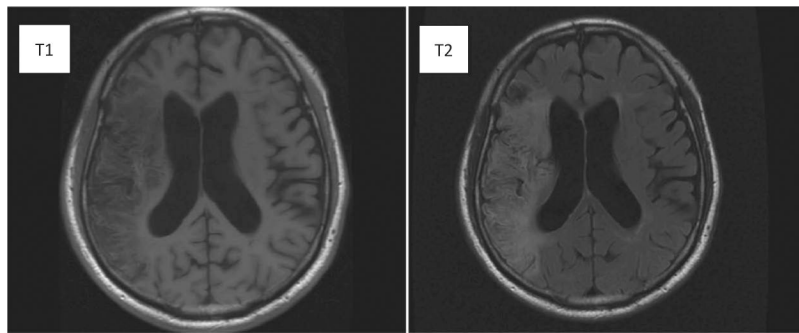


Figure 1.
Magnetic Resonance Imaging of A.R.'s Stroke.

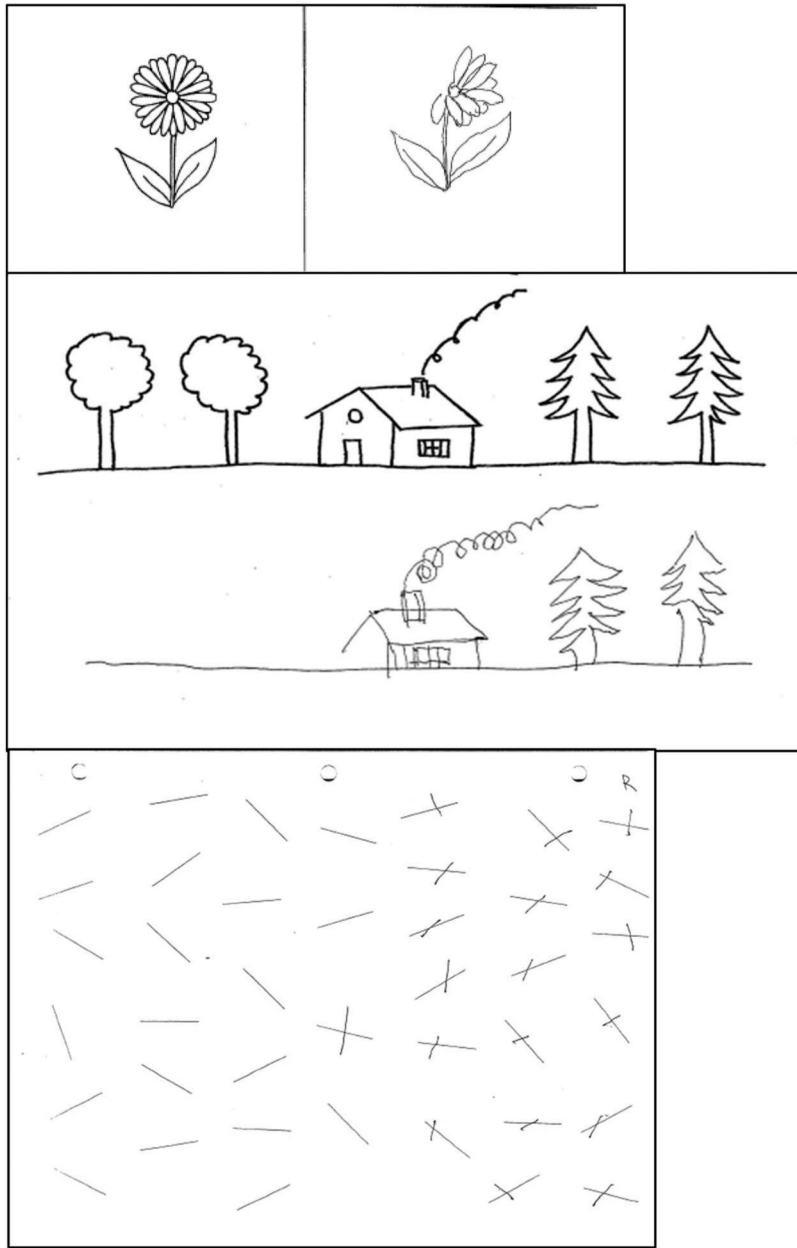


Figure 2. Drawings that required copying or actively-based Where perceptual-attentional spatial bias.

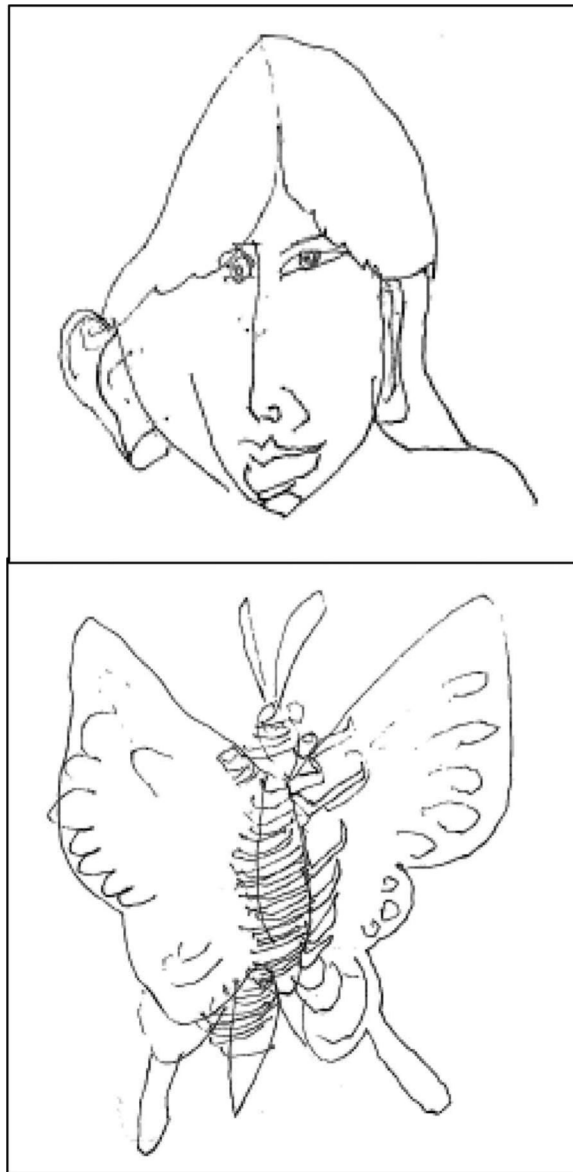


Figure 3.
Drawings completed from memory.