Retained ability to extract gist in childhood-acquired amnesia: Insights from a single case

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Abstract

Research as to whether amnesic people are able to extract gist in a converging semantic associates paradigm, known as the DRM paradigm, has produced inconsistent results in the literature. For the first time, this paper presents the performance of a young amnesic person (CJ) in this task, who acquired his memory disorder at the age of 11 years and was tested four years post-injury. In contrast with much of the data in the adult amnesia literature, CJ was found to be sensitive to the DRM manipulation at a level comparable to controls in recognition and at a level higher than controls in free recall. In addition, a detailed analysis of recall intrusions other than the critical word lent further support to the main finding that CJ is able to extract gist on the basis of semantic associations. Results are discussed with reference to the fuzzy-trace theory, the associative-activation theory and the activation-monitoring framework of false memories, as well as the potential role of an impaired and immature cognitive system in adopting a semantic gist strategy in the absence of episodic memory.

Keywords: DRM paradigm; false memories; activation-monitoring framework; amnesia; gist memory

Introduction

Gist memory refers to our ability to form and retain a conceptual or meaning-based representation of information. A prominent laboratory demonstration of gist extraction is a converging semantic associates paradigm known as the DRM paradigm (Deese, 1959; Roediger & McDermott, 1995). In a typical DRM task, participants are presented with a list of words that are strong associates of a non-presented 'critical' word. For example, participants may be presented with words such as *bed*, *rest*, *awake*, *tired*, *dream* etc., but the critical word on which the list words converge (i.e. *sleep*) is omitted from presentation. After studying a series of such lists, memory for the presented items is typically tested in recognition, or in both free recall and recognition. It has been repeatedly found that intrusion of the non-presented critical word at test (i.e. false recognition or false recall) is extraordinarily high in healthy adults (e.g. Payne, Elie, Blackwell, & Neuschatz, 1996; Roediger & McDermott, 1995; Stadler, Roediger, & McDermott, 1999). Moreover, approximately by age 11 typically developing children perform very similarly to adults in this task (e.g. Brainerd, Reyna, & Forrest, 2002; Dewhurst and Robinson, 2004; Howe, Cicchetti, Toth, & Cerrito, 2004). Due to the robustness of findings in healthy populations, the DRM effect has been understood as a 'normal distortion' of memory, which demonstrates a) our ability to extract gist from the presented information by successfully processing the semantic relationships amongst them during study, and b) that such processing occurs at the cost of generating false memories.

Three though not mutually exclusive, theories have been proposed to explain the processes involved in the DRM effect. The *fuzzy-trace theory* postulates that memory for studied items is based on two types of representations that form in parallel during study (Brainerd & Reyna, 1998). A *gist representation* is built up from associative information and contains the general meaning and interpretations conveyed during the study phase, whereas a *verbatim representation* preserves item-specific information about studied items. While both representations support veridical memory, they have the opposite effects on false memory at test: gist retrieval enhances it, verbatim retrieval inhibits it. Therefore, when participants are tested on tasks that promote the formation of a strong gist representation, such as the DRM procedure, their overreliance on gist will lead them to falsely accept test items that match this representation. The *activation-monitoring framework* on the other hand (e.g. Roediger & McDermott, 2000; Roediger, Watson, McDermott, & Gallo, 2001) emphasizes a two-stage process involving cognitive control: the critical word may be activated through semantic associations (either consciously or unconsciously) during study, which in turn creates a source-monitoring problem at test whereby participants have to distinguish between presented items and those activated mentally. Finally, a prominent theory in the developmental literature, the *associative-activation theory* proposes that the DRM effect is a product of associative-activation processes between list items and the critical word, and crucially, there is no requirement for a separate gist trace activation in order for false memories to occur (Howe, Wimmer, Gagnon, & Plumpton, 2009). Further, the automaticity with which children activate associations in their semantic memory shows a developmental trend such that automaticity increases as they get older, and so does their false memory output (Howe et al, 2009).

The performance of adult amnesic people in the DRM paradigm has been of particular interest for some time, though findings are inconsistent. Several well-documented observations about the pattern of memory impairments in amnesia lead to the hypothesis that these patients would be just as susceptible, if not more susceptible, to the DRM effect as people with intact memory. First, amnesic patients have been shown to be sensitive to pre-experimental semantic manipulations. In paired-associates learning paradigms, for instance, learning of strongly related word pairs is often better preserved than that of unrelated word pairs (e.g. Cutting, 1978; Shimamura & Squire, 1984; Winocur & Weiskrantz, 1976). Second, amnesic people tend to make more false positive errors in recognition tests than healthy controls (e.g. Knowlton & Squire, 1995; Verfaellie & Treadwell, 1993), and also often intrude more words in free recall (e.g. Schnider et al., 1996). Yet, contrary to expectations, DRM experiments carried out with adult amnesic patients (including Korsakoff amnesic people, amnesic patients with mixed etiologies and patients with dementia of the Alzheimer type) have typically reported reduced rates of false recognition of the critical word (e.g. Hudon et al., 2006; Schacter, Verfaellie, & Pradere, 1996a; Schacter, Verfaellie, & Anes, 1997; Schacter, Verfaellie, Anes, & Racine, 1998; Verfaellie, Schacter, & Cook, 2002). These findings occurred in the context of overall reduced hit rates for studied items but increased false recognition for other distracters. Based on this pattern, Schacter et al. (1996a) suggested that the formation of semantic gist representation depends on the same medial temporal lobe structures that also underlie veridical memory. More specifically, they argued that it is amnesic patients' reduced capacity to retain studied items that impedes the formation of a gist representation, ultimately causing reduced false memory for critical words in the DRM paradigm. In other words, as Melo et al. (1999, p. 344) noted, *when few, if any, of the associates (true targets) are remembered, as in the case of amnesia, there is effectively no medial temporal lobe output that can be prone to distortion* [therefore] *there is little opportunity for a related test item to match a semantic gist representation*.

Indeed, it has been demonstrated that increasing amnesic patients' veridical memory by multiple exposure to DRM lists can lead to a corresponding increase in gist-based false recognition across trials (Schacter et al., 1998). As predicted by the fuzzy-trace theory, this manipulation had the opposite effect on controls, whereby trial by trial increase in veridical memory corresponded with decreasing levels of false recognition of the critical word (see also Benjamin, 2001). Similar findings were reported more recently by Nissan, Abrahams and Della Sala (2013) in a case of amnesia, DA. DA was sensitive to the DRM effect in recognition memory to the same level as controls if veridical memory was increased by bringing her to a learning criterion of 50% correct recall on each list prior to the recognition test. Taken together, these studies strongly suggest that there needs to be a threshold level 'true memory' output in order for the DRM effect to occur, and therefore, in tests of recognition, matching veridical memory performance between patients and controls is essential to make meaningful inferences about amnesic patients' ability to extract gist.

Besides, to our knowledge, only a few studies used immediate free recall (i.e. after each study list) as a means of testing the DRM effect and the ability to form gist in amnesia. Unfortunately, Nissan et al. (2013) did not report whether or not their procedure (noted above) had led to critical word or other extra-task intrusions. In studies by Schacter et al. (1996a) and Melo et al. (1999), amnesic patients' critical word intrusion was comparable to controls, but the findings did not receive much attention as evidence of gist memory because amnesic participants also produced significantly more extra-task intrusions, thus weakening the DRM effect when adjusted measures were considered. Nevertheless, it is noteworthy that in Schacter's (1996a) study, extra-task intrusions produced by amnesic adults were predominantly semantically related to lists, which likely reflects gist memory. By our view, such observations warrant closer examination of the recall output. Therefore, in the present study we will return this issue with a careful examination of recall data to offer further insights about gist processing in the case reported here.

To the best of our knowledge, no previous work has considered the DRM effect in developmental cases of memory impairment. Here, for the first time, we report the performance of a young patient, CJ, in the DRM task. This work was motivated by the above-outlined inconsistencies in the literature as to whether amnesic patients are able to extract gist in this task. Having demonstrated strengths in semantic memory in the context of profound episodic memory and executive function difficulties (see neuropsychological profile), CJ's performance can be particularly informative to theories of gist memory in amnesia. With detailed analyses of free recall and recognition performance, we sought to offer additional insights into the DRM effect from a developmental perspective, and the conditions under which amnesic patients may be able to form and/or maintain a gist representation in this task. Special attention was paid to extra-task intrusions which are often overlooked in studies of amnesia using the DRM paradigm. A few adjustments have been made in the procedure to accommodate CJ's profound memory impairments as well as to enable meaningful inferences. Informed by previous studies, steps were taken to match recognition memory performance between CJ and controls, which are described in detail later.

The profile of CJ's retained abilities with relative strengths in semantic memory and short-term retention of information (see neuropsychological profile), and the design characteristics of the DRM paradigm may contribute to effective processing of semantic relationships between study items, and the formation of a gist representation that is accessible for at least a short period of time. Therefore, it was predicted that CJ would most likely be able to demonstrate the DRM effect under conditions of immediate free recall and possibly recognition by producing the critical non-presented word at a rate comparable to or higher than controls.

Case report

At the time of this study, CJ was a 15-year-old childhood brain tumor survivor whose case history has been reported elsewhere in detail (see Pauly-Takacs, Moulin & Estlin, 2011, 2012; Pauly-Takacs & Moulin, 2017). In brief, he was diagnosed with and successfully treated for a metastasized primary suprasellar germinoma at the age of 11 years. Figure 1 shows complete remission from cancer, but a resultant marked and generalized cerebral atrophy and associated white matter loss post treatment. Bilateral volume loss to the hippocampus was also noted in post-treatment clinical MRI scan reports.

Insert Figure 1 about here

CJ's behavior, as described by his parents and observed by us, was consistent with a profound amnesic condition. For example, he was disoriented in time and place and needed full parental support to schedule and carry out routine daily tasks. He was unable to maintain a record of ongoing activities and could not give a coherent account of what happened the day before or even earlier in the day. CJ was almost never able to answer questions about his personal past without substantial cueing, and even so, he often commented that he just “worked it out” rather than remembered events.

CJ returned to mainstream secondary education after his treatment was completed but he was only able to achieve average to good marks in subjects that he had substantial prior knowledge of. Initially he outperformed his peers on a general knowledge test. By contrast, the acquisition of novel concepts and terminology especially in newly introduced subjects became a significant challenge for him ultimately leading to a decline in his school performance. Due to his severely compromised navigation skills, he needed 1:1 support to find his way around at school at all times. Despite his extensive brain injury, there was no indication of significant language impairment; CJ’s spontaneous speech was fluent, syntactically correct and he was able to communicate effectively. CJ was also able to learn people’s names and novel facts with sufficient repetition. The impression that CJ left us with is that of a friendly and ingenious young man who had very little awareness of the profound nature and consequences of his memory deficit.

Neuropsychological Profile

Neuropsychological assessments were carried out successively, once CJ had recovered from his acute illness, at the age of 13 to 15. His overall neuropsychological profile and his everyday behavior post treatment reflects a profound anterograde amnesia with additional executive function difficulties (Table 1).

Insert Table 1 about here

As such, CJ does not present with a pure case of amnesia, but his neuropsychological profile is evidently characterized by a discrepancy between verbal IQ (taking the Wechsler Intelligence Scale for Children (WISC-IV) Verbal Comprehension Index as a proxy measure; Wechsler, 2003) and episodic memory (Children's Memory Scale (CMS) Indices; Cohen, 1997). Notably, some of the subtests pertaining to language function are in the above average range (e.g. word reading and spelling; Wechsler Individual Achievement Test (WIAT-II); Wechsler, 2005). Similarly, his scores are in the high average range on the British Picture Vocabulary test (BPVS-II; Dunn & Dunn, 1997) to suggest preserved semantic memory and relatively high premorbid intelligence. By contrast, CJ has shown virtually no episodic memory in standardized tests. For example, in the *stories* task of the Children's Memory Scale CJ was able to remember the general gist of the story, but had great difficulty retrieving specific details both immediately and after a 30-minute delay.

With respect to tests of working memory, the temporary storage of information is less affected in CJ relative to the ability of storing and manipulating information simultaneously, suggesting some residual normality in the short-term storage component of this memory system. CJ was capable of achieving within normal limits performance on *forward digit span*, but he performed consistently poorly on *backward digit span* and *letter-number sequencing* tasks yielding a poor overall Working Memory Index (Wechsler Intelligence Scale for Children (WISC-IV); Wechsler, 2003).

CJ also shows signs of a significant executive function deficit which exacerbates his memory difficulties. He experiences difficulties with *task switching*, *inhibition*, *planning and organizing behavior* as measured by the Delis-Kaplan Executive Function System test battery (D-KEFS; Delis, Kaplan & Kramer, 2001). He shows a disproportionate impairment on the *category switching* task suggesting that his main impairment is in cognitive flexibility and not in verbal fluency per se. He did not make any perseverative errors, but he tended to produce distal repetition errors. These suggest problems with *inhibitory control* and *monitoring,* which were also evident from his frequent set-loss errors as well as his low scores on the *color-word interference* task.

In sum, CJ's neuropsychological profile shows a notable dissociation between episodic and semantic memory in the context of complex cognitive difficulties: as in amnesia, he has a profound episodic memory disorder while verbal skills that draw on semantic memory are relatively well preserved. This finding is consistent with his behavior in the real world such that CJ is able to use language effectively in everyday situations. On an anecdotal note, CJ enjoyed completing verbal tasks as part of neuropsychological assessments and commented that he was "good with words". Indeed, it is CJ's well-preserved verbal skills that permitted extensive experimental work into his residual memory abilities using verbal materials, as in the task reported here.

Method

Control participants

Ten control participants (7 male) matched on years of education were drawn from CJ's class in a mainstream school. The mean age of controls was 13.07 years (SD=0.48). All participants, including CJ, and their parents signed written consent to participate in the study, and in the case of controls, for the test session to take place at school (CJ was tested at home). The study was approved by the Ethics Committee of the Institute of Psychological Sciences at the University of Leeds.

Materials and Procedure

Twelve 12-item semantically associated word-lists were adapted from previously published materials which produced a robust DRM effect in adolescents or adults (Carneiro, Albuquerque, Fernandez and Esteves, 2007; Roediger & McDermott, 1995; see Appendix 1 for the lists used). In a few instances the associates were replaced by another word to adjust to British English (e.g. *pavement* instead of *sidewalk*), or because the new word seemed more likely to elicit the critical word (*bite* instead of *canine*). In any instances of replacement, the Birkbeck word association norms were used as reference (Moss & Older, 1996).

A 72-item recognition test was designed to include 36 studied words (targets) and 36 non-studied words (distracters). The target words were those presented in positions 1, 5 and 12 of each of the previously seen lists. The 36 distracters comprised the 12 critical words as well as 24 other words which were not associated semantically to any of the study lists or to each other. The unrelated distracters were carefully selected from other DRM lists not used in this study. Similarly to the study by Roediger and McDermott (1995), the recognition test was of a blocked design whereby each block corresponded to a studied list. Each block comprised six items; three studied items, two unrelated distracters and the critical word. Each block began with a target item and ended with the critical word, the remaining items were arranged in a random order in between. The order of the blocks corresponded with the order in which the lists were studied.

In keeping with CJ's profound memory impairment and difficulty to remain focused on lengthy tasks, the task was administered to him over two sessions, one week apart. That is, in each session he was tested on six lists only, while control participants were administered the full set of 12 lists in one session. Words were presented individually on a computer screen at a rate of 2 sec per word. Participants were instructed to read the words out loud and to try to memorize them for a later test. Once they finished reading the first list, they were asked to recall as many words as they could. They were instructed that they could recall the words in any order and that they should try not to guess. Participants were given approximately 2 minutes for recall. Following this, presentation of the next list began, to be followed by an immediate free recall test as described above. This procedure repeated with subsequent lists until the recognition test was administered.

Recognition data was collected across two tests in a blocked design. CJ was tested after the third and the sixth list with each test comprising 18 items. To keep the structure of the procedure constant between CJ and controls, control participants were also tested on the first and the second half of the materials separately (i.e. after the sixth and the twelfth list) such that each recognition test comprised 36 items. The motivation behind testing CJ on fewer lists was to attempt to match recognition accuracy to controls in the standard component of the task (i.e. discounting responses for the critical distracters). To reduce the impact of short-term memory, all participants were engaged in a brief (approximately 2 minutes) conversation break immediately preceding a recognition test. Test items were read to the participants and they were asked to indicate with a yes or no answer whether the word was part of either of the lists they studied earlier. Study-test blocks were counterbalanced in the control group such that half of the participants were presented with lists 1-6 before the first recognition test, while the other half started with lists 7-12. For controls the task took approximately 40 minutes to complete, whereas CJ's sessions lasted approximately 20 minutes each.

Results

Recall

The recall data was analyzed using the cumulative recall score obtained across the 12 word lists. Our analysis strategy was to compare CJ's performance to controls with a modified t-test developed for single case studies (Crawford & Garthwaite, 2002). The mean proportion of studied and critical words recalled by CJ and controls is shown in Figure 2. CJ recalled significantly fewer studied words than did controls, t_{mod} (9) = -2.45, p = .04, z_{CC} = -2.34. However, for the non-presented critical word the inverse pattern was observed such that CJ recalled a much higher proportion of critical words than did controls. More specifically, he recalled the critical word for nine out of the 12 lists which resulted in a significantly higher false recall rate relative to controls, t_{mod} (9) = 2.44, p = .04, z_{CC} = 2.59.

Insert Figure 2 about here

Extra task intrusions

Despite the instruction to avoid guessing, intrusions other than the critical word also occurred in the output of both CJ and controls. On average, CJ produced 1.40 other intrusions per list, which was significantly higher than the average number of intrusions produced by controls, (M = 0.25, SD = 0.12), t_{mod} (9) = 9.30, p < .0001, z_{CC} = 9.58. To determine the frequency of intrusions relative to the total words recalled in the task, proportional scores were calculated for both critical words and other intrusions. These measures showed that critical and other word intrusions only minimally contributed to control participants' total recall output; with 4% and 3% respectively. By contrast,10% and 19% of CJ's total output was made up of critical words and other intrusions, respectively. Thus, his proportional intrusion rates were significantly higher for both critical word intrusions, t_{mod} (9) = 2.86, p = .02, z_{CC} = 2.92, and other intrusions, t_{mod} (9) = 7.63, p < .0001, z_{CC} = 9.55, when compared to controls.

In light of the observed differences between CJ and controls with regard to the number of intrusions produced, an adjusted measure of critical recall was calculated for each participant. To take the number of other intrusions into account, critical word recall was expressed as a proportion of the total number of intrusions (critical words + other intrusions). According to this correction, CJ intruded a numerically smaller but non-significant proportion of critical words (.35) than control participants (M = 0.52, SD = 0.23), t_{mod} (9) = -0.71, p = .50, z_{CC} = -0.74. Taken together, these analyses caution that CJ's generally greater tendency to produce intrusions may have contributed to his extraordinarily high recall of critical words (see Figure 2.). Nevertheless, we contend that the corrected recall score for critical words should not automatically be regarded as a more valid measure of gist memory. Despite CJ's greater tendency for intrusions in general, the finding that he recalled significantly more critical words than controls, cannot be ignored. Furthermore, intrusions other than the critical word may also reflect gist-based false memories if they are strongly semantically related to the lists. This possibility will be examined next.

At face value all of CJ's other intrusions appeared semantically related to their respective lists. Therefore, to obtain a finer measure, our analyses concerned the extent to which intrusions were semantically related to lists. Neither CJ nor controls intruded studied words from previous lists. Seven independent expert raters (blind to the purpose of the study and the source of intrusions) rated all critical words and all other intrusions produced by CJ and controls; they were asked to judge, on a seven-point scale, how well a given word fit the respective DRM list. The results showed that the critical words received the highest overall rating (M = 6.82, SD = 0.47) confirming that the DRM lists were successfully adapted. The second highest overall rating was given to CJ's other intrusions (M = 5.50, SD = 1.74), while control participants' intrusions received a lower overall rating (M = 4.20, SD = 2.41). An item by item analysis performed on the semantic relatedness ratings for CJ and controls showed that across all other intrusions the ratings given for CJ's intrusions were significantly higher than those given for controls' intrusions, t (293.79) = 6.62, p < .001, suggesting that CJ's intrusions were more semantically related to the lists presented than those produced by control participants. To elucidate these results, some example intrusions with their semantic relatedness ratings are provided. The three lists for which CJ did not recall the critical word he produced intrusions such as *collie* (for *dog*; 5.86), *sandstone, clay* (for *stone*; 5.43 and 5.14), *clarinet, harmony* (for *music*; 6.57 and 6.71). For the list where the critical word was *tooth*, he falsely recalled the words *molar*, *canine* and *incisor*, all of which received semantic relatedness ratings of 6.50 and higher. The lowest ratings were given to two intrusions, *encyclopedia* (3.29) and *dictionary* (3.43) produced for the list converging on *book*. Other relatively low ratings include *ugly* (4.29) and *autumn* (4.71) for lists converging on *face* and *tree*, respectively. These examples demonstrate that CJ often falsely recalled category exemplars or other strong associates of the non-presented critical word which suggests that his intrusions are supported by semantic gist.

Recognition

The recognition data was analyzed using participants' cumulative performance across multiple recognition tests. As can be seen in Table 2, CJ's proportional hit rate to true targets was equal to the mean proportional hit rate obtained by controls, t_{mod} (9) = 0. False recognition of unrelated distracters was relatively low for both CJ and the control group with CJ producing numerically more, but not significantly different number of false alarms for this type of distracter, t_{mod} (9) = 1.91, p = .09, z_{CC} = 2.29. To obtain a corrected measure of true recognition, false alarm rates to unrelated distracters were subtracted from hit rates to true targets (Budson, Desikan, Daffner, & Schacter, 2001). Comparison of CJ's and controls' adjusted hit rate revealed no significant difference between scores, t_{mod} (9) = -0.82, p = .43, z_{CC} = -0.86, which demonstrates that recognition performance on the standard component of the task (i.e. discounting responses to critical words) was successfully matched between CJ and controls.

Insert Table 2 about here

Of crucial importance is the proportion of critical distracters relative to the proportion of unrelated distracters endorsed by CJ and controls. To obtain an unbiased measure of gist memory, the proportion of unrelated distracters was subtracted from the proportion of critical distracters (Brainerd & Reyna, 1998; Budson et al., 2001). As shown in Table 2, both CJ and controls displayed great differences between false recognition of critical and unrelated distracters. A paired samples t-test confirmed that control participants' false recognition was significantly higher for critical than unrelated distracters, t (9) = 7.73, p < .001, d = 3.18, which suggests that their high rate of false recognition for critical words (i.e. 61%) was not due to response bias but was sign of a genuine DRM effect. CJ's false recognition of critical words was at ceiling (i.e. he endorsed all 12 items). However, one of our control participants also endorsed all 12 critical words (range in controls 2 to 12 words), and false recognition of the critical word was high in the control group in general (*Mdn* = 7.5; *Mode* = 6, 9). As a result, the modified t-test did not find any difference between CJ and controls, $t_{mod}(9) = 1.49$, $p = .17$, $z_{CC} = 1.60$. Overall, CJ's gist memory (i.e. adjusted false recognition) was within normal limits, $t_{mod}(9) = 1.31$, $p = .22$, $z_{CC} = 1.38$. To conclude, both CJ and control participants show a genuine DRM effect whereby high rate of false recognition of critical items cannot be explained by a generally liberal criterion for accepting test items as previously seen.

Discussion

For the first time in the literature, we presented the performance of an adolescent patient with a profound memory impairment in the DRM task. The aim of this work was to further elucidate amnesic patients' ability to form and maintain gist based on semantic associative processes whilst offering insights from a developmental perspective. CJ clearly demonstrated the DRM effect in recognition at a rate comparable to controls, and uniquely in studies carried out with amnesic people, he also demonstrated a strong DRM effect in recall at a rate significantly higher than controls. We will start by discussing the recognition findings, followed by a more detailed discussion of recall results.

Replicating previous findings carried out with similar age groups (Carnerio et al., 2007; Metzger et al., 2008), both CJ's and control participants' memory was highly vulnerable to the DRM manipulation in tests of recognition. Thus, even though CJ produced false alarms to all critical distracters, his performance was not significantly different from his healthy peers. High rates of false recognition for critical words were obtained in the context of high veridical recognition and relatively low false positives for unrelated distracters in the case of both CJ and controls, consistent with the view that gist-based false memory depends on sufficient encoding of semantic associates (Schacter et al., 1998). The normality of CJ's performance in this recognition task resembles those with developmental amnesia (DA), a memory disorder resulting from early and selective bilateral hippocampal pathology (Gadian et al., 2000; Vargha-Khadem et al, 1997), where preserved recognition memory (but impaired recall) has been confirmed as a characteristic feature of the condition (Adlam, Malloy, Mishkin, & Vargha-Khadem, 2009; Baddeley, Vargha-Khadem, & Mishkin, 2001). It is important to note however, that the high degree of similarity between the performance of CJ and controls was achieved by an adjustment in procedure which reduced memory load and the delay between CJ's first encounter with the DRM lists and the recognition test. The purpose of this modification was to attempt to increase CJ's veridical recognition performance and potentially match that with controls - at least proportionally - in order to derive meaningful inferences about CJ's ability to form a gist representation. A very low level of corrected veridical recognition memory score may mistakenly lead to underestimating the patient's ability to rely on a gist representation. Our data suggests that CJ performs similarly to controls in that he is able to derive the gist under conditions where memory load and the delay between study and test are optimized for his episodic memory deficit. Our findings are consistent with other patient work where modifications in procedure ensured that there is sufficient item-specific memory output from which to extract gist (Schacter et al., 1998; Nissan et al., 2013).

CJ's high recognition accuracy suggests that task characteristics inherent in the DRM paradigm are particularly beneficial for his memory. More specifically, a converging semantic associates paradigm promoted the formation of a strong gist representation during study which supported not only his decisions to accept items that matched this representation but also to reject those that did not. In other words, being able to rely on gist, CJ was able to adopt a relatively conservative response criterion. However, as predicted by the fuzzy-trace theory (Brainerd & Reyna, 1998), not being able to rely on a strong verbatim trace, this response criterion was not conservative enough to suppress false recognition of the critical word.

CJ's performance was strikingly different from that of controls for immediate free recall. He recalled significantly fewer studied items but, as predicted, significantly more non-presented critical words compared to his healthy peers. By showing marked differences in this direction, CJ's performance pattern is unique amongst studies carried out with people with significant memory impairment. CJ's strong tendency to falsely recall the critical word suggests that he is able to extract the semantic gist in a list learning task and use this information to guide his recall. Additional analyses of other extra-task intrusions revealed that although CJ had a greater tendency than controls to intrude non-presented words, these were not arbitrary. Rather, they were semantically related to the respective DRM lists providing further evidence that recall was driven by gist. In light of the finding that typically developing 11-year-old children (i.e. CJ's age at injury) make more semantic intrusions than other types of recall error in the DRM task (Dewhurst & Robinson, 2004), CJ's performance demonstrates a particularly high degree of susceptibility to gist-based false memories in a developmental context. CJ's strong tendency to intrude semantically related words into his recall can also be accommodated by the associative activation theory (Howe et al 2009); by this account, CJ activated associations in his well-retained semantic knowledge base rather like healthy adults (i.e. automatically) ultimately leading to an increased level of false memories.

There are several factors that were likely to contribute to the particular pattern of CJ's recall performance, which will be discussed in turn. Given the immediacy of recall, the contribution of short-term memory is evident (c.f. Van Damme and d'Ydewalle, 2009a), although short-term memory capacity alone does not explain the processes involved in gist extraction and subsequent false recall. We propose that CJ's performance may be a consequence of severe episodic memory loss combined with monitoring difficulties subserved by frontal regions of the brain. It is well established that the prefrontal cortex plays a crucial role in monitoring and verifying the output from memory (e.g. Moscovitch & Melo, 1997). By this account, the impoverished verbatim trace (i.e. impaired veridical recall) may have forced CJ to rely on the gist trace which he successfully formed during study, ultimately leading him to mentally generate items which matched this representation, including the critical word and other gist-relevant words. In addition to this, deficient monitoring of gist-related information further contributed to an unverified recall output characterized by a high proportion of false memories. Indeed, a striking finding in CJ's recall output was that all intrusions were semantically related to their respective list, more so than intrusions produced by control participants. This lends further support to the main finding of this report: CJ's retrieval in the DRM task is strongly driven by gist.

Melo et al. (1999) have attempted to delineate the influence of temporal and frontal damage alone or in combination on false memory in the DRM paradigm. They have found that those patients whose amnesia was complicated with frontal deficits (i.e. functionally most comparable to CJ) were not sensitive to the DRM effect in either recall or recognition. However, patients with a pure form of amnesia appeared more prone than controls to intrude the critical word into recall, although the difference was not significant. Thus, curiously, CJ's performance in the DRM task is more similar to those adults with amnesia who do not have additional frontal difficulties, despite the fact that such neuropsychological difficulties are clearly present. Melo et al. (1999) proposed that the reason why those patients with combined medial-temporal and frontal deficit did not display the DRM effect is mainly due to the fact that they were unable to derive the gist or use it effectively because of poor strategic processing. That is, in their view, strategic processing is required for extracting gist in the DRM paradigm. The data obtained from CJ is in contrast with this proposal and suggests that it is possible to successfully extract and rely on gist representation (at least in this paradigm) despite severe impairments in episodic memory and a range of executive functions. We suggest that age at injury might account for this difference, however, in the absence of studies using the DRM paradigm in childhood-onset amnesia it is not possible to fully appreciate this possibility. Further to this, neuropsychological evidence suggests that the ability to extract gist from semantic associates and the ability to monitor the output from memory do not rely on the same areas of the frontal cortex. Schacter et al. (1996b) reported a case (BG), whose injury affected the lateral regions of the right frontal lobe and has shown a striking over-reliance on memory for the general characteristics of a study episode, but was unable to effectively monitor related, specific information. Another patient (JB), who had a putative lesion to the prefrontal cortex, experienced the added difficulty of being unable to extract gist (Parkin, Bindschaedler, Harsent, & Metzler, 1996). More recently Warren et al. (2014) found that damage to the ventromedial prefrontal cortex also led to a reduction in the DRM effect. What can be concluded from this is that CJ's diffuse brain injury has either spared the prefrontal cortex sufficiently enough for him to be able to extract gist in the DRM paradigm, or alternatively, extracting gist from semantically associated words is less reliant on strategic processing than thought.

In fact, studies carried out with Korsakoff amnesic patients are in support of this latter suggestion. Van Damme and d'Ydewalle (2009b; 2010) reported that Korsakoff amnesic patients' veridical and false memory scores were diminished only in explicit memory tests, but not when memory was tested implicitly. Employing a 'think aloud' protocol, Van Damme and d'Ydewalle (2010) found that even though patients verbalized fewer critical words than controls during study, they showed normal priming for these words. This led the authors to conclude that amnesic patients do not need to consciously activate the critical words in order to extract gist in the DRM paradigm.

One of the earliest ideas to explain false memory phenomena in the context of verbal learning also emphasizes the importance of implicit associations. The *implicit associative response* (IAR) theory postulates that false memories arise because semantic associates of the presented words become activated during study through automatically spreading activations (Underwood, 1965). Underwood does not rule out the possibility that sometimes the activated word reaches conscious awareness: *It must be clear that IAR, in most* *theoretical formulations, is conceived of as actually occurring. This is to say, it is not a hypothetical construct. It is hypothetical only in the sense that it is assumed to occur in a particular situation where it cannot be observed directly, and this assumption is made because it has been observed to occur overtly with a certain frequency in other situations* (Underwood, 1965, p. 122). An anecdotal note from our work illustrates this point. CJ enjoyed completing this task, and despite no reference made to the fact that the words to be presented would be related to each other in some way, he often commented on the lists during presentation, for example: "These words all have to do with *sleep*!" He was keen to discover the theme of a list during study, and in doing so, he activated the critical non-presented word by overt verbalization for three of the lists. He also anticipated that all lists will have a theme, and often asked what the next set of words "would be about". These observations confirm that CJ engaged with semantic processing to form a gist representation during study and lend direct support to the activation-monitoring theory of false memories (Roediger & McDermott, 1995; 2000); CJ activated critical words during study and endorsed them at test as a result of committing reality monitoring errors. Conscious activation of the critical lure at study, as observed in CJ, typically strengthens the illusion (e.g. Seamon et al., 2002), unless participants engage in a specific form of retrieval monitoring whereby, they recall-to-reject the critical lure, which in turn leads to decreased susceptibility to the DRM effect. Such monitoring often occurs when participants are explicitly warned of the illusion (Neuschatz, Benoit, & Payne, 2003), but can also occur without warning (Carneiro, Fernandez, & Dias, 2009). Relevant to our study, Carneiro et al. found age-related effects in whether or not participants apply a recall-to-reject strategy as a function of identifiability of the critical lure. For highly identifiable critical distracters adults employed a recall-to-reject strategy resulting in a reduced DRM effect, but children and pre-adolescents (i.e. 11-12-year olds) produced more false memories for such distracters. In light of these findings, CJ's high rate of free recall of the critical word demonstrates a particularly high degree of DRM susceptibility relative to controls. His performance is beyond what could be explained by normative immaturity of monitoring abilities and is therefore most likely a consequence of his brain injury affecting prefrontal regions. Related to this, we have shown elsewhere that CJ has particular difficulty with adopting the recall-to-reject strategy even if there is sufficient source information available to support such decisions (Pauly-Takacs & Moulin, 2017).

Limitations and future directions

In summary, the present paper furthers our understanding of the DRM effect in amnesia and the neuropsychological circumstances under which gist extraction may occur. CJ's performance demonstrated sensitivity to the DRM illusion with particularly high susceptibility in free recall conditions; a result not typically found in the adult amnesia literature. Our results suggest that neither profound episodic memory difficulties nor deficits in executive function necessarily impede semantic associative activation processes to extract and maintain gist. Our patient's DRM susceptibility is best explained by well-retained semantic memory processes coupled with poor metacognitive monitoring of the activated memory output.

Due to the diffuse nature of CJ's brain injury acquired in childhood, and the unavailability of detailed neuroanatomical data we were unable to relate findings to specific brain regions implicated in the component processes of the DRM effect. Rather, our approach was to provide a detailed analysis of performance and contextualized this with existing theories and neuropsychological findings concerning the phenomenon. One possibility is that CJ's unique pattern of brain injury and cognitive profile predisposed him to enhanced gist processing. Alternatively, profound episodic and executive deficits acquired in the context of the developing brain might exacerbate cognitive weaknesses (i.e. monitoring) whilst leaving other functions, such as semantic memory, intact. CJ's case demonstrates that gist extraction may become habitual by way of adapting to life without episodic memory as an adolescent. Indeed, it was found elsewhere that CJ adopts a semantic gist strategy when attempting to retrieve daily life events, and he does so without prompting (Pauly-Takacs, Moulin & Estlin, 2011). Further support for this possibility comes from studies carried out in developmental amnesia which have demonstrated that not only do DA patients retain the capacity to learn new semantic information (e.g. Baddeley et al, 2001; Gardiner, Brandt, Baddeley, Vargha-Khadem, & Mishkin, 2008; Guillery-Girard, Martins, Parisot-Carbuccia, & Eustache, 2004; for a review see Elward & Vargha-Khadem, 2018), but may even use semantic gist to boost episodic-like recall (Brandt, Gardiner, Vargha-Khadem, Baddeley, & Mishkin, 2006). It may be the case that semantic memory mechanisms are more available in developing cognitive systems in active learning environments, especially when episodic memory is compromised, which would explain the differences in DRM task performance between CJ and those that acquired amnesia in adulthood. A fruitful avenue of research would be to continue to examine the cognitive mechanism and brain bases underlying gist extraction in brain injury, especially of developmental age. In light of recent work suggesting that false memories induced by the DRM task primed solutions to insight-based verbal problems in both children and adults (Howe, Garner, Charlesworth, & Knott, 2011; Howe, Threadgold, Norbury, Garner, & Ball, 2013), it would be particularly interesting to explore the adaptive nature and potential rehabilitation utility of strong false memory tendencies in other patients and patient groups whose deficit is similar to that of the case reported here. Childhood brain tumor survivors like CJ are an emerging population in neuropsychology, often presenting with significant episodic memory and executive function difficulties (Nagel et al., 2006; Conklin, Ashford, Howarth, & Merchant, 2012), thus the need for a better understanding of the relative strengths and weaknesses in cognition to inform rehabilitation potential is warranted.

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Disclosure of interest

The authors report no conflict of interest.

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Appendix 1

Word lists used in the DRM task

tree	***book***	***face***	***stone***	***bread***	***music***	***rain***	***bed***	***car***	***dog***	***door***	***tooth***
leaves	read	eyes	hard	butter	note	water	sleep	wheel	puppy	knob	cavity
fruits	pages	nose	rock	food	sound	umbrella	sheets	engine	cat	lock	dentist
nature	letters	mouth	floor	eat	piano	drops	pillow	steering-wheel	animal	house	mouth
log	school	freckles	land	sandwich	sing	cloud	mattress	van	bark	wood	white
root	study	look	nature	rye	radio	wet	blanket	bonnet	leash	key	bite
flowers	reading	ear	throw	jam	band	weather	cosy	ride	collar	gate	gum
forest	stories	wash	pebble	milk	melody	cold	bedroom	accident	friend	entrance	chew
wood	sheets	beautiful	big	flour	horn	thunderstorm	sleepy	drive	bite	open	rotten
plant	cover	person	granite	jelly	concert	winter	dreams	road	kennel	bell	paste
shade	pen	head	pavement	dough	instrument	sun	lay	Ferrari	hair	window	brush
green	pencil	skin	mason	crust	symphony	liquid	chair	tires	doghouse	close	braces
branch	magazine	smile	heavy	slice	jazz	wind	rest	transport	tail	latch	wash

The word in bold at the top of each list is the critical (non-presented) word for that list.

Figure 1

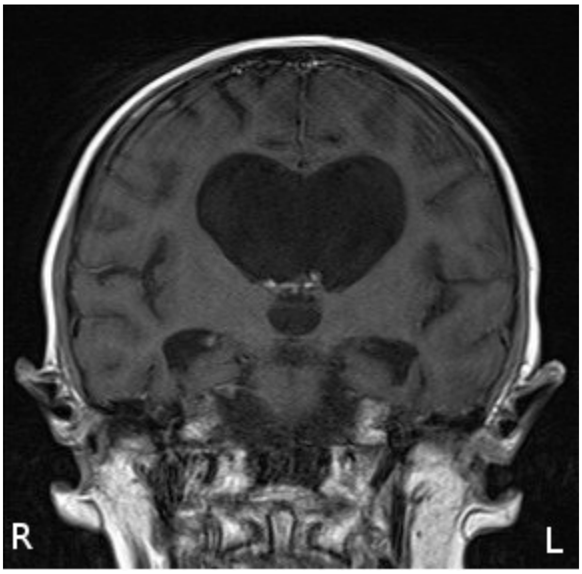

Figure 2

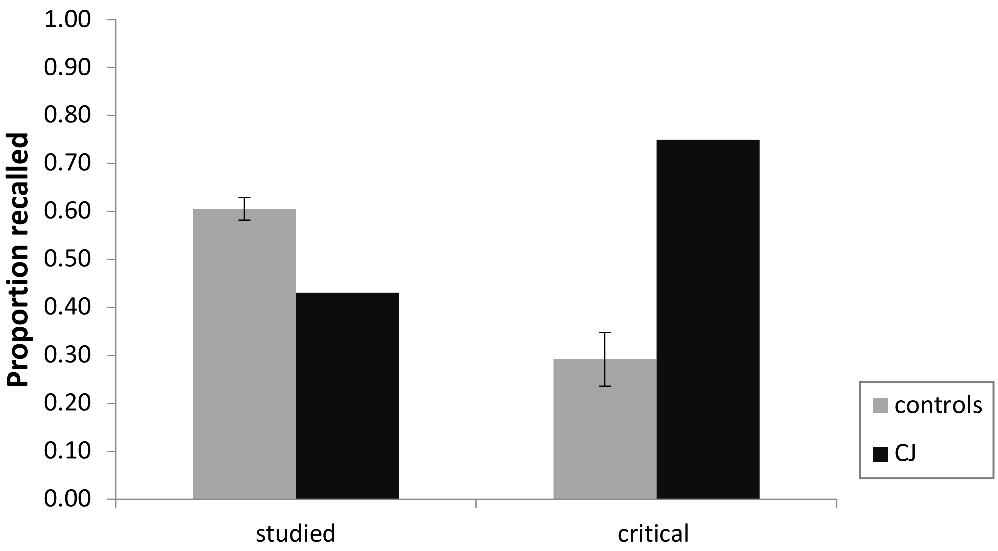
Proportion recalled
1.00
0.90
0.80
0.70
0.60
0.50
0.40
0.30
0.20
0.10
0.00
studied
critical
controls
CJ

Table 1: Neuropsychological profile of CJ

Test	Scaled score	Percentile Rank/ Comments
General Cognitive Functioning		
WISC-IV Subtests		
Performance Indices		
Verbal comprehension	96	39 (average)
Perceptual reasoning	57	0.2
Working memory	54	0.1
Processing speed	50	0.1
Premorbid functioning		
BPVS		
Receptive vocabulary	109	72 (high average)
WIAT-II		
Word reading	118	(high average)
Reading comprehension	48	<0.1
Pseudoword decoding	102	55
Numerical operations	59	0.3
Mathematical reasoning	56	0.2
Spelling	114	(high average)
Written expression	n/c	
Listening comprehension	82	12
Oral expression	74	4
Memory		
CMS Performance Indices		

Visual immediate memory	50	< 0.1
Visual delayed memory	50	< 0.1
Verbal immediate memory	54	0.1
Verbal delayed memory	54	0.1
Attention and concentration	82	12
Learning	50	< 0.1
Delayed recognition	66	1
Executive Functioning		
D-KEFS		
Verbal fluency		
Letter fluency (total correct)	8	(low average)
Category fluency (total correct)	9	(average)
Category switching (total correct)	1	(severely impaired)
Percent set-loss errors	1	(severely impaired)
Percent repetition errors	1	(severely impaired)
Color-word interference		
Completion times		
Color naming	7	(low average)
Word-reading	7	(low average)
Inhibition	2	(severely impaired)
Inhibition / Switching	3	(impaired)
Error analysis		
Inhibition	3	(impaired)
Inhibition / Switching	1	(severely impaired)
Tower Test		
Total achievement score	1	(severely impaired)

Notes: WISC-IV = Wechsler Intelligence Scale for Children – Fourth Edition; BPVS = British Picture Vocabulary Scale; WIAT-II = Wechsler Individual Achievement Test – Second Edition; CMS = Children's Memory Scale; D-KEFS = Delis – Kaplan Executive Function System

Table 2: Proportion of items judged 'old' in the DRM task by CJ and controls, and the adjusted recognition memory accuracy scores.

	CJ	Controls
Targets	.86	.86 (.07)
Critical distracters	1.00	.61 (.25)
Unrelated distracters	.08	.02 (.03)
Adjusted true recognition	.78	.84 (.07)
Adjusted false recognition	.92	.59 (.24)

Note: Values in brackets represent one standard deviation of the mean.

Figure captions

Figure 1: CJ's brain following treatment with chemotherapy and radiotherapy. Marked and generalized cerebral atrophy and loss of hippocampal volume remain (coronal T1 weighted sequence with gadolinium).

Figure 2: Proportion of studied and critical non-presented words recalled by CJ and controls. Error bars represent one standard error of the mean.