

Prospective, declarative, and nondeclarative memory in young adults with spina bifida

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Abstract

The consequences of congenital brain disorders for adult cognitive function are poorly understood. We studied different forms of memory in 29 young adults with spina bifida meningocele (SBM), a common and severely disabling neural tube defect. Nondeclarative and semantic memory functions were intact. Working memory was intact with low maintenance and manipulation requirements, but impaired on tasks demanding high information maintenance or manipulation load. Prospective memory for intentions to be executed in the future was impaired. Immediate and delayed episodic memory were poor. Memory deficits were exacerbated by an increased number of lifetime shunt revisions, a marker for unstable hydrocephalus. Memory status was positively correlated with functional independence, an important component of quality of life. (*JINS*, 2007, *13*, 312–323.)

Keywords: Hydrocephalus, Neural tube defects, Immediate memory, Delayed memory, Recall, Recognition

INTRODUCTION

Spina bifida meningocele (SBM), a neural tube defect that is the product of a complex pattern of gene–environment interactions, is associated with distinctive physical, neural, and cognitive phenotypes (Dennis et al., 2006a; Fletcher et al., 2004). Improvements in medical and surgical techniques (in particular, management of hydrocephalus with shunt technologies developed in the 1960s; Blum et al., 1991) have produced a cohort of individuals with SBM who now range in age from young adulthood to middle age, and whose cognitive function is only now being studied (e.g., Dennis & Barnes, 2002; Hetherington et al., 2006; Huber-Okraïnec et al., 2002).

Individuals with SBM have lesions at various levels of the spinal cord, and a higher spinal lesion level is associ-

ated with more brain compromise and greater cognitive morbidity in children (Fletcher et al., 2005). In adults with SBM, spinal lesion level affects motor skills such as speech production (Huber-Okraïnec et al., 2002) and motor independence (Hetherington et al., 2006), although not cognitive–academic function (Dennis & Barnes, 2002; Hetherington et al., 2006).

Another source of variability in individuals with SBM is the number of lifetime shunt revisions, a marker for the stability of hydrocephalus. Cognitive outcome is generally unrelated to number of childhood shunt revisions (Jensen, 1987; Raimondi & Soare, 1974; Ralph et al., 2000; Tromp et al., 1979; but see Halliwell et al., 1980). In adults, the number of shunt revisions is negatively related to functional numeracy, independent living, and employment (Dennis & Barnes, 2002; Hetherington et al., 2006; Hunt et al., 1999).

Children with SBM exhibit a distinctive cognitive phenotype, with variations *across* domains [e.g., higher Verbal IQ (VIQ) than Performance IQ (PIQ)]; Dennis et al., 1981;

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Fletcher et al., 1992; and better reading than mathematics; Barnes et al., 2006] and *within* domains (e.g., facility with words but deficits with texts; Dennis et al., 1994). This cognitive profile appears to persist into adulthood (e.g., young adults with SBM show reduced PIQ relative to VIQ; Hetherington et al., 2006; Hommet et al., 1999).

Memory is a key cognitive resource, both for the acquisition of cognitive and academic skills in childhood and for the maintenance of adult cognitive and academic competencies. Reviewing earlier studies (e.g., Parsons, 1969), Wills (1993) concluded that memory was not a characteristic deficit of children with SBM, although later studies suggested specific rather than global memory deficits (e.g., Scott et al., 1998; Yeates et al., 1995). The risk of memory problems increases with both normal and pathological aging (Cabeza et al., 2005). Adult survivors of SBM with hydrocephalus have poorer memory than adults with occult spina bifida (Barf et al., 2003) but the specificity of memory deficits has not been established.

Prospective memory, the recall of intentions to be activated in the future (cf. retrospective memory, the recall of past events or information), may be event-based (e.g., delivering a message when you see a particular person), time-based (e.g., remembering to keep an appointment at a specific hour), or activity-based (e.g., remembering to take a pill after dinner) (Kvavilashvili & Ellis, 1996). Prospective memory has not been studied in children with SBM, although these children can remember to apply rules in upcoming situations (Fletcher et al., 1996). Prospective memory deteriorates with age (Craik & Grady, 2002), but it is not known whether it does so more sharply in individuals with SBM than in typically developing individuals.

Declarative memory involves the conscious recall of experiences, facts, or events (cf. *nondeclarative*, or implicit memory, the alteration of performance without conscious meditation) (Loring, 1999). Children with SBM have preserved implicit memory and motor learning (Dennis et al., 2006b; Edelman et al., 2004; Yeates & Enrile, 2005) but impaired explicit memory (Yeates & Enrile, 2005).

Working memory is a limited capacity storage system in which items can be manipulated for complex tasks (Loring, 1999). Children and young adults with SBM have poor working memory spans and poor information maintenance (Dennis & Barnes, 2002; Mammarella et al., 2003; Vachha & Adams, 2005). Strategic aspects of working memory may be less impaired (e.g., when remembering lists of fruit or animal words, children with SBM can use the information that fruit gets a higher score than animals to guide recall; Vachha & Adams, 2005).

Semantic memory, the recall or recognition of context-free knowledge, appears intact in children with SBM (Vachha & Adams, 2005). Children with hydrocephalus, most with SBM, perform within the normal range on vocabulary, an expression of semantic memory (Barnes et al., 2001).

Episodic memory is the recall or recognition of context-specific events. Children with hydrocephalus, including those with SBM, have difficulties with delayed (but not immedi-

ate) recall (Scott et al., 1998; Yeates et al., 1995; reviewed in Ewing-Cobbs et al., 2003) and with list learning involving error correction (Fletcher et al., 1992). Young adults with SBM have impairments of word list learning and memory (Barf et al., 2003) and delayed spatial memory (Iddon et al., 2004).

The distinction between preserved nondeclarative memory and impairments in some forms of declarative memory may be robust with age and therefore be evident in young adults with SBM. Like children with SBM, young and middle-aged adults with SBM show age-appropriate vocabulary (Barnes et al., 2004a), one indicator that semantic memory may be intact over much of the lifespan, whereas tests of word list learning, general memory, spatial memory, and information maintenance are impaired (Barf et al., 2003; Dennis & Barnes, 2002; Iddon et al., 2004), suggesting some difficulties with episodic and working memory. Nondeclarative memory in adults with SBM has not been studied.

Cognitive competencies have been shown to affect the level of adult independence. In the same sample of adults with SBM used in the present study, Dennis and Barnes (2002) found a measure of functional math skills, but not functional literacy, to be significantly correlated with measures of social interaction and communication, personal living, and community independence.

The present study investigates memory in young adults with SBM. The first aim is to establish whether the profile of memory function in adult life is similar to or different from that reported for children with SBM. We hypothesized:

1. Because there is no evidence to indicate problems with prospective memory in children with SBM, young adults with SBM will have intact prospective memory.
2. Like children with SBM, adults with SBM will have difficulty with working memory. Specifically, they will show impairments in maintenance and manipulation of information but may not have difficulty in cognitive inhibition and set shifting.
3. Like children with SBM, adults with SBM will have intact semantic memory.
4. Because children with SBM have problems with various types of episodic memory tasks, adults with SBM will show impairments in episodic memory; specifically, intact immediate and delayed recognition and intact immediate recall, but impaired delayed recall.
5. As in children with SBM, adults with SBM will have intact nondeclarative memory.

The second aim is to evaluate memory as a function of one source of biological instability, lifetime shunt revision history. We predicted that individuals with an unstable shunt revision history would have poorer memory, just as shunt instability has been related to poor functional numeracy (Dennis & Barnes, 2002). The third aim is to relate memory to self-report indices of functional independence. We pre-

dicted that significant memory difficulties would be associated with lower levels of independence.

METHOD

Participants

Participants were 29 adults with SBM and shunted hydrocephalus (*M* age = 26.60 years; *SD* = 4.82; range = 18.50–36.23; 13 women, 16 men). Each had been treated for hydrocephalus with a diversionary shunt shortly after birth or in early infancy. Each had a VIQ or PIQ score of 70 or above on the Wechsler Adult Intelligence Scale, Revised (WAIS-R, Wechsler, 1981). Mean VIQ score was in the average range, whereas the mean PIQ score was just below 1 *SD* from the norm (*M* VIQ = 94.93; *SD* = 9.23; range = 79–114; *M* PIQ = 84.55; *SD* = 10.22; range = 70–114). Education levels varied [secondary school (11), community college/incomplete undergraduate program (15), completed undergraduate degree (2), graduate degree (1)]. Six participants had upper lesions (T12 and above, corresponding to van Allen et al.’s, 1993, closure site 1), and 23 had lower lesions (L1 and below, corresponding to van Allen et al.’s closure site 5). The number of shunt revisions (*M* = 9.40; *SD* = 18.67; range = 0–100) was used to create two groups, one whose shunt history involved five or fewer revisions (*n* = 17; *M* = 1.88; *SD* = 1.53; median = 2; range = 0–5) and one (*n* = 12; *M* = 20.04; *SD* = 25.93; median = 12.5; range = 6–100) whose shunt history involved more than five revisions. Additionally, scores from a sample of 29 healthy controls were used as a comparison to the participants with SBM (*M* age = 26.88 years; *SD* = 5.86; range = 18.21–36.58; 18 women, 11 men) for some measures (see below). Controls were siblings, relatives, and friends from a study of brain tumors. All participants gave informed consent for participation in the study, which complied with institutional ethics standards.

The SBM and control groups did not differ in age [$F(2,55) = .64, p > .05$] and a rank-sum test indicated that the groups did not differ in gender ($p > .05$). The SBM groups had lower WAIS scores than controls [$F(2,55) = 5.79, p < .01, F(2,55) = 28.62, p < .001$] for the VIQ and PIQ measures, respectively (see Table 1). Controls scored

Table 1. Age and IQ for controls (*n* = 29) and participants with spina bifida meningomyelocele (SBM) with five or fewer shunt revisions (*n* = 17) or six or more shunt revisions (*n* = 12)

	Age	VIQ	PIQ
Controls	26.88 (5.86)	103.97 (10.76)	105.03 (11.67)
SBM group:			
0–5 revisions	27.53 (4.83)	94.88 (9.33)	87.88 (10.49)
SBM group:			
>5 revisions	25.28 (4.69)	95.00 (9.50)	79.83 (8.01)

Note. VIQ = Verbal IQ; PIQ = Performance IQ.

higher than both shunt revision groups on both VIQ ($p < .05$) and PIQ ($p < .001$), but the scores from the two groups of SBM participants did not differ.

This study group was the same as (or subsumed) that in four previous reports of adults with SBM, including studies of reading and writing skill, math and numeracy, neuro-motor speech deficits, and quality of life (Barnes et al., 2004a; Dennis & Barnes, 2002; Hetherington et al., 2006; Huber-Okraimec et al., 2002).

Procedures

Participants were administered the MicroCog™ Assessment of Cognitive Functioning (MicroCog™, Powell et al., 1993), the Test of Everyday Attention (TEA, Robertson et al., 1994), and the Rivermead Behavioural Memory Test (RBMT, Wilson et al., 1985). Figure 1 shows the mapping of the measures from these tests on the constructs of memory discussed in the introduction. Parents were administered the Scales of Independent Behavior, Revised (SIB-R, Bruininks et al., 1996), a self-report measure of functional independence. The measures are described in further detail below.

MicroCog™

The MicroCog™ is a computerized test of cognition, from which we used the following subtests, each with scores expressed as age-based and education-adjusted scaled scores (*M* = 10; *SD* = 3).

Numbers Forward

In this task, which involves the maintenance component of working memory, participants reproduce visually presented

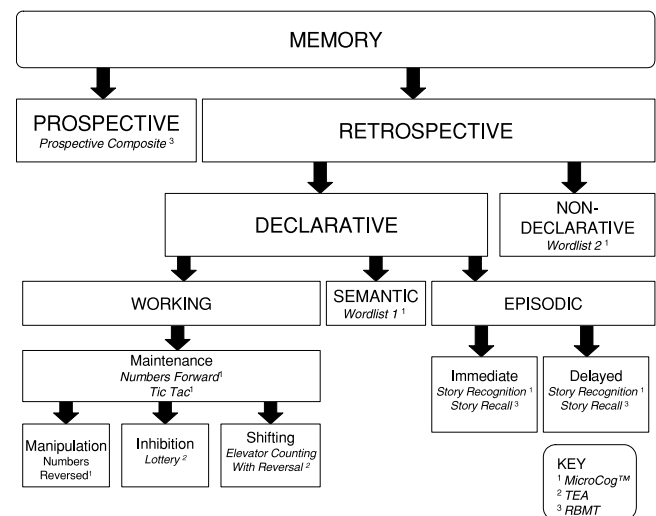


Fig. 1. The relation of the measures from the MicroCog™, the Test of Everyday Attention (TEA), and the Rivermead Behavioural Memory Test (RBMT) to the constructs of memory.

two- to nine-digit sequences on a numeric keypad. The first item has a string length of five digits (each appearing for 1 s), and digit span is increased or decreased by one depending on response accuracy or is discontinued with three consecutive failed items. Total number of correct items and average response time are recorded.

Tic Tac

This task requires the ability to maintain spatial information and to reproduce the locations of stimuli set in a grid matrix. Participants view a 1-s display of a 3×3 matrix in which three, four, or five of the nine spaces are colored, and then replicate the pattern by pressing the corresponding keys on a keypad. Two sets of images with different configurations are presented and discontinued with two failed items. The total number of correct items is summed across the two presentations, and the response times are averaged.

Numbers Reversed

In this task, which requires manipulation of information in addition to maintenance, participants reproduce visually presented digit sequences in reversed order on a numeric keypad. Digit span range and administration is the same as for Numbers Forward. The total number of correct items and average response time are recorded.

Wordlist 1

This measure of semantic memory requires participants to decide whether specific items belong to a superordinate category. A list of the same 16 words is presented four times. The first and second presentations involve decisions about two semantic categories (clothing and occupations). The third and fourth presentations involve decisions about phonemic categories beginning with the letter “s” or a vowel. The words appear one at a time for approximately 1 s each, and total score is recorded. Raw response time is averaged but not scaled.

Story (immediate) 1 and 2

This task involves immediate recognition of verbal material. Participants answer 5 (Story 1) or 6 (Story 2) multiple choice questions about each visually presented story. Each question has four response options. The total score is summed across both stories, and the response times are averaged.

Story (delayed) 1 and 2

This task involves delayed recognition of verbal material. After a filled delay of 20–30 min following the visual presentation of two brief stories, each with different content, the participant’s recognition of story details is tested by 11 (Story 1) or 12 (Story 2) multiple choice questions for each story. Questions and responses are presented in the same format as in the immediate Story subtest. Total scores and

average response times are combined for the Story and Address (see below) subtests.

Address

This task also assesses delayed recognition. After a filled delay following the visual presentation of a name and address, the participant’s recognition of these details is tested by four multiple choice questions in the same format as the Story task. Delayed recognition total score and response time are combined with the Story scores.

Wordlist 2

This task involves nondeclarative learning of a categorized word list and is presented immediately after Wordlist 1. The participants are given a list of 36 words presented for 1 s each, including the 16 words presented repeatedly in Wordlist 1 that they were not explicitly instructed to remember. Participants must respond with a keypress to indicate words that were on the previous list, and total score is recorded. Raw response time is averaged but not scaled.

Timers

This simple reaction time task requires participants to press a key as quickly as possible to a visual stimulus that appears on the screen. Five stimuli are presented in each of two separated blocks for a total of 10 trials. Trials appear from 1 to 5 s after a cue and receive a maximum score of 2 s if no response is made before then. This task was included as a control for differences in motor speed.

TEA

We used scaled scores from the following TEA working memory subtests.

Lottery

This task, which requires maintaining stimuli in working memory and inhibiting irrelevant responses, has the format of the standard N-back working memory task (Cohen et al., 1993). Participants are instructed to search for winning numbers (e.g., one that ends in 55) in a 10-min series of audio-taped numbers of the form “BC143” and to write down the two letters preceding all numbers ending in 55. There are 10 winning numbers presented among a total of 212 items at a rate of one item every 2.8 s.

Elevator Counting with Reversal

This working memory and set shifting task (Robertson et al., 1994) requires participants to pretend they are in an elevator with a nonfunctional floor indicator and to establish which floor they have arrived at by counting a series of tape-presented tones. There are 10 strings of tones, ranging in length from 4 to 18 tones, presented at a rate of one tone per second. Participants count the high and low tones, switch-

ing direction of elevator movement with a tone change. Data were available for this subtest on 25 of the SBM participants.

RBMT

The RBMT provides analogues of everyday memory situations (remembering to carry out an everyday task, or retaining newly learned information). We used raw scores from the following subtests.

Prospective Memory

This score is a composite of the scores for the Belonging, Appointment, and Message subtests testing prospective memory. In the Belonging task, participants need to remember to ask for the return of their own hidden belonging at the end of the 25-min test and remember where it was hidden. In the Appointment task, an alarm is set for 20 min and the participant is required to ask a particular question relating to the near future when it sounds. In the Message immediate and delayed (20 min) tasks, the participant must remember to drop off a message at a certain point during another subtest requiring the participant to trace a path around a room. Raw scores for these tasks are summed to yield the prospective memory score with a possible total score of 12 points, with only partial points given if the participant needs to be reminded to ask the question or deliver the message.

Story (immediate)

This task involves the immediate recall of verbal information. Participants listen to a reading of a short prose passage five to six lines in length with 21 details and recall as much detail as possible. Points and half-points are assigned for all recalled or partially recalled details and summed to yield the raw score for this task.

Story (delayed)

This task involves the delayed recall of verbal information. After a delay of approximately 20 min, participants recall the prose passage. Performance is scored as in the immediate recall condition, and one point is deducted if the participant requires a cue to recall the passage.

SIB-R

Standard scores from the SIB-R assessed the following clusters.

Social Interaction and Communication Skills

This subscale assesses social interaction, language comprehension, and language expression skills. Participants or their parents rate how well tasks such as making plans with friends, taking telephone messages, and filling out application forms are completed without assistance.

Community Living Skills

This subscale assesses time and punctuality, knowledge of money and its value, work skills, and home/community orientation. Participants or their parents rate how well tasks such as writing down appointments, paying bills, responding to criticism, and using public transportation are completed without assistance.

Data Analyses

The SBM participants were separated into groups according to number of shunt revisions for all analyses. The results from each task were analyzed in one of two ways, depending on whether control participants completed that subtest. For the RBMT, which does not yield age appropriate scaled scores, raw scores obtained from participants with SBM were compared with the results of normal controls. Because the RBMT scores were not normally distributed, Kruskal–Wallis (H) analyses were used to explore group differences.

For the standardized MicroCog™ and TEA scaled score measures, the shunt revision groups were compared with the population means with one-sample t tests. Only MicroCog™ scaled scores based on total correct, and not response time, were included in these comparisons. A Bonferroni adjustment of the significance level (with a rounded corrected value indicated in the significance statement for each significant measure) was used to control the error rate for each specific hypothesis and in *post hoc* testing of the significant Kruskal–Wallis effects. To facilitate comparisons between the different constructs of memory, effect sizes and their respective confidence intervals were generated for each subtest.

Because individuals with an upper spinal lesion are less frequent both in terms of number of live births and number of survivors, our sample was unbalanced for spinal lesion level. To investigate possible effects of lesion level, the data also were analyzed nonparametrically with all participants with SBM included to explore differences between those with an upper and lower lesion. As well, the main parametric analyses were repeated with only those individuals with a lower lesion level included to ensure the results held with the more homogeneous group of SBM participants.

Memory measures were correlated with IQ, age, and the two measures of functional independence. Differences were assessed as a function of handedness. To evaluate the effect of differences in motor speed, simple reaction time scores from the MicroCog™ Timers subtest were correlated with all MicroCog™ response time measures and with the TEA Lottery subtest, which has a motor component.

RESULTS

RBMT mean raw scores, obtained for both SBM participants and controls, are presented in Table 2. Mean scaled scores for the MicroCog™ and TEA subtests, obtained for the SBM participants only, are presented in Table 3.

Table 2. Mean raw Rivermead Behavioural Memory Test (RBMT) subtest scores for controls and participants with spina bifida meningomyelocele (SBM) by shunt revision group

Memory construct	Subtest	Controls	SBM group 0–5 revisions	SBM group > 5 revisions
Prospective	Prospective composite	11.48 (.79)	11.00 (1.23)	10.25 (1.49)*
Episodic–immediate	Story (recall)	9.48 (3.31)	7.71 (2.72)	6.33 (2.15)*
Episodic–delayed	Story (recall)	8.47 (3.30)	5.91 (2.64)*	4.71 (2.50)*

*Significantly different from controls ($p < .05$) with Bonferroni adjustment.

Prospective Memory

SBM group and control participants differed in performance on the prospective memory composite [$H(2) = 7.71$, $p < .05$]. The group with more than five revisions, but not the group with fewer revisions, showed significantly poorer performance than controls ($p < .02$). The two SBM groups did not differ from each other.

Working Memory

Maintenance

On the MicroCog™ tasks requiring maintenance of presented stimuli, SBM participants' performance differed depending on the specific task and shunt revision group. Total correct scores for the group with fewer revisions, but not the group with more revisions, were significantly below the norm for the Numbers Forward subtest [$t(16) = 2.84$, $p < .01$]. However, scores for the Tic Tac subtest were significantly below the population mean for both shunt groups [$t(16) = 3.88$, $p < .01$, $t(11) = 4.73$, $p < .01$].

Manipulation

On the MicroCog™ Numbers Reversed task requiring maintenance and manipulation of presented stimuli, both shunt

groups performed more poorly than the population mean [$t(16) = 4.41$, $p < .03$, $t(11) = 2.85$, $p < .03$].

Cognitive inhibition

All participants with SBM performed within the normal range on the Lottery subtest, which requires response inhibition.

Set shifting

On the Elevator Counting with Reversal subtest, which requires participants to shift their response pattern on cue, participants with SBM in both revision groups scored comparably to the population mean.

Semantic Memory

On the MicroCog™ Wordlist 1 subtest, both groups of participants with SBM demonstrated performance comparable to the population mean.

Episodic Memory: Recognition

Immediate

On the MicroCog™ Story subtest of immediate recognition memory, participants with SBM in both revision groups performed comparably to the population mean.

Table 3. Scaled scores on the MicroCog™ and Test of Everyday Attention (TEA) subtests for participants with spina bifida meningomyelocele (SBM) by shunt revision group

Memory construct	Test battery	Subtest (normed mean = 10.00)	SBM group 0–5 revisions	SBM group > 5 revisions
WM-maintenance	MicroCog™	Numbers Forward	8.06 (2.82)*	8.50 (2.61)
	MicroCog™	Tic tac	7.65 (2.50)*	6.58 (2.50)*
WM-manipulation	MicroCog™	Numbers Reversed	8.00 (1.87)*	7.75 (2.73)*
WM-cognitive inhibition	TEA	Lottery	11.00 (3.22)	8.42 (3.92)
WM-set shifting	TEA	Elevator Counting with Reversal	8.13 (4.22)	7.20 (3.52)
Semantic	MicroCog™	Wordlist 1	9.65 (3.06)	8.00 (4.55)
Episodic-immediate	MicroCog™	Story (recognition)	8.18 (4.77)	7.42 (3.90)
Episodic-delayed	MicroCog™	Story (recognition)	7.24 (3.80)*	5.75 (3.82)*
Nondeclarative	MicroCog™	Wordlist 2	11.06 (1.09)*	10.67 (3.11)

Note. WM = working memory; MicroCog™ = MicroCog™ Assessment of Cognitive Function; TEA = Test of Everyday Attention. * $p < .05$ with Bonferroni adjustment.

Delayed

On the MicroCog™ Story and Address subtests of delayed recognition, both shunt groups scored lower than the population mean [$t(16) = 3.00, p < .03, t(11) = 3.86, p < .03$].

Episodic Memory: Recall

Immediate

SBM groups and control participants differed in performance on the RBMT Story task of immediate recall [$H(2) = 7.59, p < .05$]. Individuals in the group with more than five revisions, but not those in the group with fewer revisions, scored lower than controls ($p < .02$). The two SBM groups did not differ.

Delayed

SBM groups and control participants differed in performance on the RBMT Story task of delayed recall [$H(2) = 13.26, p < .01$]. Compared with controls, both shunt revision groups with SBM had difficulty remembering the information in the RBMT Story subtest after a delay ($p < .02$), although the two shunt groups did not differ from each other.

Nondeclarative Memory

On the MicroCog™ Wordlist 2 subtest, participants with SBM and fewer than six shunt revisions, but not those with more revisions, scored higher than the population mean [$t(16) = 4.01, p < .03$].

Memory Profile

Figure 2 displays effect sizes with the associated 95% confidence intervals for each memory measure. Confidence intervals are reported because effect size estimates based on smaller samples are more variable, despite the fact that effect size computations are independent of sample size. Participants with SBM performed at comparable levels to the norm on the semantic and nondeclarative memory tasks and showed more variable performance on the memory tasks assessing working and episodic memory. Effect sizes of the level of impairment were moderate to large ($>.50$) for RBMT prospective memory (significantly so only for the group with more revisions) and working memory maintenance and manipulation (MicroCog™-Numbers Forward, Tic Tac, Numbers Reversed), but were small for a memory task involving inhibition (TEA-Lottery). The working memory task involving set shifting (TEA-Elevator Counting with Reversal) and the semantic memory task (MicroCog™-Wordlist 1) yielded moderate to large effect sizes especially for the group with more revisions, but these differences were nonsignificant due to high variability. Effect sizes were moderate to large for immediate and delayed recall and recognition (RBMT-Story, MicroCog™-Story), although the

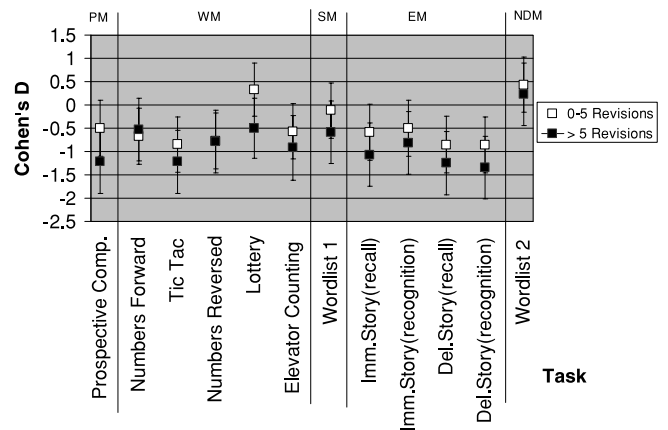


Fig. 2. Effect sizes and their associated 95% confidence intervals for performance by participants with spina bifida meningomyelocele (SBM) across the different constructs of memory. Performance by participants with SBM was compared with performance by controls for the Rivermead Behavioural Memory Test (RBMT) measures and to normative standards for the MicroCog™ and Test of Everyday Attention (TEA) measures. PM = prospective memory; WM = working memory; SM = semantic memory; EM = episodic memory; NDM = nondeclarative memory; Imm. = immediate; Del. = delayed.

impairment on the immediate tasks only attained significance in one case. The nondeclarative memory task (MicroCog™-Wordlist 2) generated small effect sizes.

Supplementary Analyses: Memory and Spinal Lesion Level, VIQ, Age, and Motor Speed

Upper and lower spinal lesion level groups with shunt revision groups combined did not differ in performance on any of the memory measures when compared by nonparametric analyses ($p > .05$). As well, when the performance on the RBMT tasks by participants in the lower lesion level group only was compared with controls, the prospective memory and the immediate and delayed Story subtest scores were significantly different between groups ($p < .05$). The control group scored significantly higher than the group with lower lesions and more shunt revisions, but not those with fewer shunt revisions, on all three measures ($p < .02$). Comparisons for the participants with lower lesions only to the normed scores for each subtest from the MicroCog™ and TEA generally yielded the same pattern of impairments as with all participants combined.

Most memory scores were uncorrelated with VIQ. VIQ was significantly correlated with the RBMT Story subtest for both immediate recall [$r(27) = .41, p < .05$] and delayed recall [$r(27) = .47, p < .05$], but the difference between controls and the SBM group with more revisions remained significant for delayed recall only when VIQ was included as a covariate [$F(2,54) = 3.39, p < .05$]. VIQ was significantly correlated with the TEA Elevator Counting with

Reversal task [$r(23) = .47, p < .05$] and the MicroCog™ Numbers Forward task [$r(27) = .40, p < .05$], which is not surprising because repeating digits tasks are included in both the WAIS-R and the MicroCog™. The pattern of tasks that are performed poorly by participants with SBM is not a simple reflection of those tasks that are correlated with IQ, nor are memory tasks performed poorly across the board in this sample of participants with SBM.

To assess whether there are age effects on the RBMT, which is not standardized according to age and education levels, separate correlations were performed between age in months at RBMT assessment and performance on this task for the SBM and control groups. No correlation was significant in either group.

Reaction time correlated significantly with response time on only two measures, the MicroCog™ Tic Tac scaled score [$r(27) = -.40, p < .05$] and the Wordlist 1 raw score [$r(27) = .39, p < .05$].

Relations Between Functional Independence and Memory

The mean standard score on the Social Interaction and Communication Skills cluster was within the normal range ($n = 29$; $M = 103.14$; $SD = 18.25$). Scores on this scale correlated positively with the TEA Elevator Counting with Reversal score [$r(23) = .56, p < .01$] and delayed recall on the RBMT Story subtest [$r(27) = .42, p < .05$].

The mean standard score on the Community Living Skills cluster was within the normal range ($n = 28$, $M = 100.21$, $SD = 24.22$). Scores on this scale correlated positively with the MicroCog™ Tic Tac score [$r(26) = .40, p < .05$] and the TEA Elevator Counting with Reversal score [$r(22) = .50, p < .05$].

DISCUSSION

Young adults with SBM exhibited a variable memory profile. Prospective memory was poor with a less stable hydrocephalus history, contrary to our hypothesis. As predicted, nondeclarative memory and semantic memory were intact, whereas aspects of episodic declarative memory were impaired. Hypotheses about working memory were confirmed, with the profile showing good working memory requiring response inhibition and rule application but poor working memory with high information maintenance and input transformations. Overall, the findings suggest a developmentally stable memory profile in individuals with SBM from childhood to young adulthood, with adults showing additional problems in prospective memory and components of episodic memory.

The data bear on several more general issues: the dissociation between nondeclarative and declarative memory, preserved semantic memory and impaired episodic memory, the selectivity of working memory deficits, the role of shunt instability in memory deficits, the putative neural basis of

memory impairments, and memory deficits and quality of life.

Our broadest functional dissociation was that between intact nondeclarative memory and impaired declarative memory. Consistent with these data, children with SBM show intact perceptual and conceptual priming in implicit memory tasks (Yeates & Enrile, 2005), but variable declarative memory (Scott et al., 1998; Yeates & Enrile, 2005). Learning shows an analogous dissociation to memory, namely, intact procedural learning but impaired overt performance. Impaired motor performance in children and adolescents with SBM (Hetherington & Dennis, 1999; Salman et al., 2005) coexists with intact motor adaptation for saccades (Salman et al., 2006), ballistic arm movements (Dennis et al., 2006b), and prismatic distortion (Colvin et al., 2003), and with intact motor sequence learning (Edelstein et al., 2004). Dissociations between intact implicit learning and impaired explicit performance have been demonstrated both across (Colvin et al., 2003; Grimm, 1976; Hetherington & Dennis, 1999) and within (Edelstein et al., 2004; Salman et al., 2005, 2006) groups. SBM does not appear to compromise the development of nondeclarative, implicit, and adaptive memory and learning.

Semantic memory is preserved in young adults with SBM, just as in children with this condition, who acquire lexicon and vocabulary (Barnes & Dennis, 1992; Barnes et al., 2001; Horn et al., 1985). Young adults with SBM also show age-appropriate vocabulary, suggesting that core lexical representations and semantic memory may be intact over much of the lifespan (Barnes et al., 2004a).

Children with SBM have intact immediate episodic recall (Scott et al., 1998), whereas some adults with SBM demonstrated intact immediate recognition but impaired immediate recall. Both typically aging adults and children with SBM show poorer episodic memory on recall tests than on tests of recognition (Craik & McDowd, 1987; Yeates et al., in press). In contrast, the delayed episodic memory deficits in young adults with SBM were independent of the assessment method (recognition vs. recall).

Both children and adults with SBM have selective rather than global impairments of working memory. In adults with SBM, working memory problems may have consequences for on-line language comprehension, because understanding sentences and texts requires active working memory (Clifton & Duffy, 2001; van den Broek et al., 1999). Sentence and text comprehension is impaired in children with hydrocephalus, most with SBM (e.g., Dennis & Barnes, 1993). Comprehension deficits are driven, at least in part, by problems holding semantic information in working memory, because these children are more disadvantaged than controls by greater textual distance between sentences to be integrated (Barnes et al., 2004b).

Children and adolescents with SBM have well-developed attention skills requiring top-down cognitive control, such as the ability to attend to endogenously cued, cognitively interesting information (Dennis et al., 2005a,b). Consistent with these data, young adults with SBM were able to per-

form working memory tasks with low working memory load even if required to inhibit irrelevant material on the basis of a top-down rule or set shift from one rule to another. However, they had difficulty with working memory tests requiring input manipulations.

Less stable hydrocephalus, indexed by a larger number of shunt revisions, was associated with poorer memory in several domains. Hydrocephalus is the final common path of several conditions ranging from spina bifida to perinatal intraventricular hemorrhage, tumors, and adult normal pressure hydrocephalus with dementia. Hydrocephalus causes structural and functional anomalies of the hippocampus. Animal models of hydrocephalus show macroscopic structural integrity of the hippocampus, but dendritic, axonal, and synaptic alterations suggest deafferentation of dark pyramidal neurons (Kriebel & McAllister, 2000). Furthermore, impairment of spatial memory in kaolin-induced hydrocephalic rats is associated with dysfunction of the hippocampal cholinergic and noradrenergic systems (Egawa et al., 2002). The structure of the hippocampus in adults with SBM has not been reported, and the data on the brains of young adults with SBM is limited to two structural studies (Hommet et al., 2002; Rand-Hendriksen & Christensen, 1998) that essentially describe the brain stigmata of Chiari II evident in children with SBM (Fletcher et al., 2005).

Even if well controlled, hydrocephalus contributes to poor memory. Although individuals with more shunt revisions had poorer prospective and immediate episodic memory, our young adults with SBM and fewer revisions still showed memory impairments. Young adults with SBM who have nonfunctioning shunts or nonshunted ventriculomegaly show improved memory after shunt treatment (Mataro et al., 2000). On the other hand, Hommet et al. (2002) found no relation between memory and magnetic resonance imaging (MRI) ventricular dilatation (a measure of concurrent hydrocephalus) in a small sample of average-intelligence young adults with hydrocephalus.

Although hydrocephalus is common to several groups with brain insult, and may exacerbate memory deficits, memory profiles vary with etiology of the hydrocephalus and, presumably, with the distinct patterns of brain damage associated with different etiologies. Hommet et al. (1999) found that young adults with SBM had poorer memory than those with a different congenital etiology, aqueduct stenosis. A study of memory in children and adolescents with treated brain tumors of the third and fourth ventricle and variable hydrocephalus found implicit as well as explicit memory deficits (Dennis et al., 1998).

Memory function was positively correlated with communication and community living independence. Power limitations precluded exploration of the relation between functional independence and memory using path analyses to delineate causal patterns, a task that remains for future studies. Nevertheless, the correlation of memory with functional independence suggests that the needs of young adults with SBM may include the provision of memory thera-

pies, retraining, or support at an earlier point in life than the age at which independence-limiting disorders of memory would typically emerge. To live independently, individuals need to remember prospective and retrospective events on a daily basis, such as class times, prescription pick-ups, and passwords for bankcards. Because limited independence likely translates into limited academic achievement and job opportunities (Dennis & Barnes, 2002), overall quality of life is likely affected in these individuals (see Hetherington et al., 2006, for measures of quality of life in this sample).

A limitation of the present study is that control participants were not tested on all memory measures. The results of the comparisons to normative data are only true under the assumption that this sample is comparable to the normative sample. Additionally, longitudinal comparison of child and adult memory could not be made because there are no theoretically meaningful childhood memory scores on our study participants. In a study of physical, medical, educational, and employment status in young adults with SBM, late deterioration in several domains has been reported (Bowman et al., 2001), which may include functional memory. On the basis of the number and extent of memory deficits in adults with SBM, including prospective memory impairments, one would advance the hypothesis that memory in adult life is more widely impaired than in childhood, at least for individuals with an unstable shunt revision history.

Our adult participants are still relatively young, the oldest being 36 years of age. To be sure, the age range for studies of neurocognitive function in adults with SBM in other studies is even more limited (in Hommet et al., 1999, the oldest participants were in their mid-twenties). Our current spina bifida project investigates memory in adults with SBM up to 55 years of age. Following the first MRI studies in young adults with SBM (e.g., Hommet et al., 2002), we will soon be able to relate MRI morphometric and diffusion tensor imaging measures to multiple measures of basic memory constructs, brain changes with aging, shunt history, and chronological age, and to provide tests of the hypothesis that memory in individuals with SBM is one of several physical and neurocognitive functions that show greater compromise with increasing age and medical instability.

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