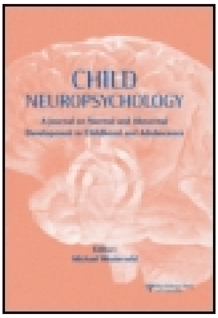

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Selective and sustained attention in children with spina bifida myelomeningocele

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Selective and sustained attention in children with spina bifida myelomeningocele

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Spina bifida myelomeningocele (SBM) is a neural tube defect that has been related to deficits in several cognitive domains including attention. Attention function in children with SBM has often been studied using tasks that are confounded by complex motor demands or tasks that do not clearly distinguish perceptual from response-related components of attention. We used a verbal-report paradigm based on the Theory of Visual Attention (Bundesen, 1990) and a new continuous performance test, the Dual Attention to Response Task (Dockree et al., 2006), for measuring parameters of selective and sustained attention in 6 children with SBM and 18 healthy control children. The two tasks had minimal motor demands, were functionally specific and were sensitive to minor deficits. As a group, the children with SBM were significantly less efficient at filtering out irrelevant stimuli. Moreover, they exhibited frequent failures of sustained attention and response control in terms of omission errors, premature responses, and prolonged inhibition responses. All 6 children with SBM showed deficits in one or more parameters of attention; for example, three patients had elevated visual perception thresholds, but large individual variation was evident in their performance patterns, which highlights the relevance of an effective case-based assessment method in this patient group. Overall, the study demonstrates the strengths of a new testing approach for evaluating attention function in children with SBM.

Keywords: Distractibility; Impulsivity; Lapses; Perception thresholds; TVA; Visual processing speed.

Spina bifida myelomeningocele (SBM) is a congenital neural tube defect. It is the most common form of spina bifida; a condition that has a postdietary-fortification prevalence of about 0.3–0.5 per 1,000 live births in the United States (Fletcher, Dennis, & Northrup, 2010). This makes neural tube malformations one of the most common types of birth defect (Refsum, 2001; Yoon et al., 2001). SBM, which is often associated with hydrocephalus, involves a complex pattern of neural damage and functional impairments. SBM-related brain insults are diffuse and affect children from embryonic stage and throughout life in orthopaedic, cognitive, and behavioral domains (Burmeister et al., 2005; Fletcher et al.,

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2004; Wills, 1993). The neurobehavioral profile associated with myelomeningocele and hydrocephalus has been reviewed by several authors (e.g., Dennis & Barnes, 2010; Fletcher et al., 2005; Liptak, 2002). SBM is associated with impaired movements in upper and lower limbs (Fletcher et al., 1995, 2004; Hetherington & Dennis, 1999; Lomax-Bream, Barnes, Copeland, Taylor, & Landry, 2007) and consequently reductions in complex fine motor skills related to eye-hand coordination, for example, writing (Barnes, Dennis, & Hetherington, 2004; Ziviani, Hayes, & Chant, 1990) and drawing (Sandler, Macias, & Brown, 1993). The observable deficits may reflect impairments in motor timing as well as spatial perception and construction guided by vision (Dennis, Landry, Barnes, & Fletcher, 2006; Fletcher, Francis, Thompson, Davidson, & Miner, 1992; Liptak, 2002; Lomax-Bream et al., 2007; Vinck, Mullaart, Rotteveel, & Maassen, 2009). Typical cognitive outcomes involve deficits in multiple mental domains including timing, attention, perception, memory, literacy, numeracy, and language functions (Dennis & Barnes, 2010).

Attention function in individuals with SBM has been investigated in relatively few studies (Yeates, Fletcher, & Dennis, 2008) but is potentially an area of central interest. In general, attention plays the role of a selection mechanism that prioritizes cognitive resources and allows only the most relevant information to influence consciousness and action (Bundesen & Habekost, 2008). Attention is therefore fundamental to appropriate behavior, learning, and development. Based on parent-rating scales assessing behavioral criteria for attention deficit/hyperactivity disorder (ADHD) according to the *Diagnostic and Statistical Manual of Mental Disorders*, fourth edition (*DSM-IV*, American Psychiatric Association, 2000), it has been estimated that one fourth of children with SBM exhibit difficulties in this area (Burmeister et al., 2005; Fletcher et al., 2005; Rose & Holmbeck, 2007). Attention problems in children with SBM are also suggested by reports of higher prevalence of ADHD (Ammerman et al., 1998; Burmeister et al., 2005; Fletcher et al., 2005; Gadow & Sprafkin, 1987; Swartwout et al., 2008). In these studies the majority of children with ADHD symptoms were classified as belonging to the predominantly inattentive subtype, which is characterized by distractibility and poor sustained attention. It should, however, be noted that these estimates were based on rating scales designed to screen for ADHD rather than fully diagnose the condition. Other studies have used experimentally based methods to describe attention function in SBM. Dennis et al. (2005a, 2005b) used covert orienting paradigms to examine attentional orienting in children and adolescents with SBM. The results revealed several significant tendencies: Children with SBM showed slowed covert orienting to both exogenous and endogenous cues. Moreover, mean reaction times (RTs) reflected a larger disengagement cost for exogenously cued targets in the vertical plane cued at short intervals (200 ms) and an attenuated inhibition of return at long intervals between cue and stimulus (1000 ms). Thus, children with SBM in this study exhibited a number of specific, not global, deficits of attentional orienting. Also, impairments in sustained attention have been found in children with SBM using different versions of continuous performance tests (CPTs). Main dependent variables in CPTs include false alarm responses (commission errors), taken to reflect poor response inhibition, and missing responses (omission errors), taken to reflect lapses of attention. Several studies have found significantly more errors of commission and omission in children with SBM (Brewer, Fletcher, Hiscock, & Davidson, 2001; Lollar, 1990; Loss, Yeates, & Enrile, 1998; Swartwout et al., 2008). In order to estimate the children's ability to maintain an alert state over time, Swartwout et al. included an analysis of performance stability across the duration of the CPT. Children with SBM showed consistent performance across the testing session without significant changes in reaction time, and Swartwout et al. concluded that the ability to sustain attention does not differentiate children with SBM from typically developing controls.

Several studies have found that executive functions, which may be regarded as higher order attentional control processes, are impaired in children and adolescents with SBM (Brewer et al., 2001; Gioia, Isquith, Guy, & Kenworthy, 2000; Loss et al., 1998; Mahone, Zabel, Levey, Verda, & Kinsman, 2002; Rose & Holmbeck, 2007; Snow, 1999). However, Fletcher et al. (1996) suggested that the impaired performance on executive tasks is not attributable to classic frontal dysfunction. Cognitive measures from children with SBM on the Wisconsin Card Sorting Test, the Stroop Color and Word Test, as well as the Tower of London were instead interpreted to reflect impairments in basic attention and motor functions (alertness and processing speed). In agreement with Fletcher et al. (1996), processing resources were found to be reduced in a study by Jacobs, Northam, and Anderson (2001), who showed significantly impaired performance among children with SBM on the Coding subtest of the *Wechsler Intelligence Scale for Children,* third edition (*WISC-III*) and The Rey-Osterrieth Complex Figure Test (as measured by time of completion).

Reviewing the literature on children with SBM, it is evident that attention research is of large relevance. However, the research field seems to have room for improvement in at least three areas. First, performance on some attentional tasks is potentially confounded by impairments of fine motor ability, eye-hand coordination, and visuospatial cognition. This is especially the case for tasks that require speeded complex motor responses (e.g., writing, drawing, or visually guided manipulation of elements, as required in The Trail Making A and B tests, the Coding and Symbol Search subtests of the WISC-III, and The Tower of London). Though many studies have used tasks with more simple motor demands, the need for data that are less contaminated by deviant motor function is generally acknowledged in the SBM literature (e.g., Fletcher et al., 1996; Loss et al., 1998; Swartwout et al., 2008; Vinck et al., 2009, 2010). Vinck et al. (2009, 2010) have recently addressed this specific need. They found children with SBM to perform normally on computerized reaction time measures of sustained, focused, and divided attention, attentional flexibility, and impulsivity and concluded that tasks with simple motor requirements (press of a response button) offered good validity to SBM studies. To our knowledge, however, attention function in children with SBM still remains to be investigated without the aid of reaction time measures.

A second limitation of the existing literature is a lack of functionally specific and theoretically based measures, especially concerning the nonmotor aspects of attention. Dennis et al. (2005a, 2005b) used the attention model of Posner (1980) to analyze data from orienting paradigms and to measure specific processes like inhibition of return and the speed of covert orienting. However, perceptually defined, nonmotor aspects of attention such as visual distractibility, spatial encoding bias, and visual processing speed have not previously been measured and should also be hard to disentangle from RT-based data due to the significant response component in the measurements. Furthermore, the studies of Dennis and collaborators did not include performance measures related to sustained attention.

Third, many children with SBM may have subtle attention deficits that go undetected in traditional tests. For example in relation to sustained attention, target and nontarget stimuli in CPTs are usually presented in random order. However, it has been argued that this unpredictability may have an alerting effect in itself, and if stimuli are always presented in the same, fixed sequence, response behavior is more likely to become automatic and vulnerable to attention lapses in the way everyday behavior can be (Dockree et al., 2006). The sensitivity to attentional lapses may also be enhanced by embedding a secondary task in the CPT that further distracts the participant and challenges the fronto-parietal networks responsible for sustaining attention (Dockree et al., 2006).

The present study has two main aims: (a) to introduce a stronger methodology for research on attention deficits in children with SBM and (b) to use these methods to map individual profiles of attentional function. Motivated by the methodological challenges described above, we chose to examine a group of children with SBM using two attention tests: (a) a letter-identification experiment (combined whole and partial report) based on the Theory of Visual Attention (TVA; Bundesen, 1990) and (b) the Dual Attention to Response Task (DART) developed by Dockree et al. (2006). Both tasks are well established in studies of adult normal and clinical populations, but new to the SBM literature. Neither of the two tasks should be affected by the motor impairments related to SBM: Performance on the TVA-based paradigm depends solely on accuracy (unspeeded verbal reports) and effectively dissociates attention mechanisms in the visual system from motor processes. In the DART, speeded responses are limited to key presses, a simple response form that should not put children with SBM at a motor disadvantage compared to healthy children (Dennis et al., 2005a; Vinck et al., 2010).

The TVA-based test can provide measurements of attentional processes that are both functionally specific and grounded in basic attention research: The TVA model conceptualizes visual processing as a parallel processing race (Bundesen, 1990; Desimone & Duncan, 1995), in which the different stimuli in the visual field compete for representation in a short-term memory store of limited capacity. The model describes the encoding process in terms of five distinct mathematical parameters: the perception threshold, t_0, visual processing speed, C, storage capacity of visual short-term memory, K, visual distractibility, α, and the relative attentional weight (i.e., proportion of the total available processing capacity) of each visual object, w. As detailed in the section entitled "CombiTVA," these five parameters can be measured separately using a simple letter-identification task. Bundesen (1990) used the formulas of TVA to account for a wide range of classical findings on focused and divided attention, including results from whole report, partial report, single stimulus recognition, cued detection, and visual search (see Bundesen & Habekost, 2008, for an updated account of the literature). Bundesen, Habekost, and Kyllingsbaek (2005) went on to show that the same equations can account for many attentional effects at the level of single neurons in the brain. TVA has also been employed in numerous studies of adult neurological patients, where the attention parameters defined in the model have been estimated in conditions such as visual neglect (Duncan et al., 1999), simultanagnosia (Duncan et al., 2003), alexia (Starrfelt, Habekost, & Leff, 2009), and Huntington's disease (Finke et al., 2005). Because of its functional specificity and theoretical grounding, attention testing based on TVA should also be highly relevant to studies of neurological disorders in children, including SBM. Besides being functionally specific, TVA-based testing has also been shown to be very sensitive to minor attention problems that are not detected in standard tests (Habekost & Bundesen, 2003; Habekost & Rostrup, 2006; Habekost & Starrfelt, 2006). The whole and partial report experiments feature very brief visual stimulation (below 200 ms) that is close to the threshold for conscious perception. As a consequence, minor abnormalities in visual processing are more likely to be revealed. No previous study has measured the attentional parameters defined by TVA in children with SBM, but based on clinical reports and the prevalence of ADHD (inattentive subtype), we hypothesized that visual distractibility (the α parameter) would be compromised in the patient group. Concerning the four other parameters of visual attention, we had no firm hypotheses and viewed the study as largely exploratory. However, for possible deviating parameters other than α, expectations were that patients would perform below the level of controls.

The TVA-based test does not provide measures of sustained attention. To investigate this aspect of attentional function we chose the DART, which should be more sensitive to sustained attention deficits than traditional CPTs for two reasons. First, there is a secondary task embedded in DART (detection of odd-colored stimuli) to distract the participant and to further challenge the ability to sustain attention. In accordance with this, Dockree et al. (2006) showed that DART provides measures of sustained attention with increased sensitivity compared to an earlier, single-task version of the test, the SART (Robertson, Manly, Andrade, Baddeley, & Yiend, 1997). Second, the fixed presentation sequence in DART imitates the predictability of monotonous everyday tasks, in which maintenance of an appropriate level of attention must be endogenously induced. This way, alertness can be measured with greater sensitivity. For these reasons, DART should provide a sensitive tool for assessing sustained attention in children with SBM. Based on the existing literature, we expected that attentional impairments in terms of both response inhibition (commission errors) and attentional lapses (omission errors) would be frequent in our patient group.

METHODS

Participants

Children with SBM were recruited from Rigshospitalet in Copenhagen, Denmark. Rigshospitalet is the regional treatment center for SBM patients in the eastern part of Denmark. Patients were selected on the basis of medical records. To be selected for participation, a patient should satisfy the following inclusion criteria: (a) age between 10 and 14 years, (b) no history of major psychiatric disease (e.g., ADHD) or specific learning disability, assessed by parent report and medical records, (c) no color blindness, (d) normal or corrected-to-normal vision, as assessed with the Snellen Chart (McGraw, Winn, & Whitaker, 1995; Peters, 1961), and (e) intellectual skills within the normal range as assessed with WISC-III (Weschler, 1992). Two potential participants were excluded from the study based on these criteria: An 11-year old girl who was diagnosed with mild mental retardation after completing all 10 WISC-III subtests (corresponding to ICD-10 code F70) and a 13-year old boy who had been diagnosed with ADHD and received medical treatment. Neuroimaging data (CT or MR scans) were available for all patients and were used to establish the presence of medical variables (a case overview is presented in Table 1). One scan (patient RD) dated close to birth, while three scans (patients JS, KI, and FA) were obtained more recently (i.e., within 3–6 years before testing). Two scans (patients LP and NK) were obtained less than 1 year before testing. The mean age of the patients was 12.7 years ($SD = 0.9$) and the group consisted of 4 girls and 2 boys.

Eighteen neurologically healthy control subjects formed an age-matched control group (15 girls and 3 boys, mean age of 12.5 years, $SD = 1.0$). The control group did not differ significantly from the patient group in terms of age or gender (age: Mann-Whitney $U = 49$, $p = .7$; gender: Fishers Exact Test; $p = .568$, both tests were two-sided).

The control children were recruited from schools in the local area. Exclusion criteria were a history of psychiatric disease, specific learning disabilities, or color blindness. All controls had normal or corrected-to-normal vision, assessed with the Snellen Chart, and all were right-handed. For every patient and control subject, a parent or legal guardian gave informed written consent prior to participation according to the Helsinki Declaration. The WISC-III (Weschler, 1992) was administered to both groups, and eight subtests were chosen on the basis of the short form presented by Donders (1997), who suggested these

Table 1 Medical and Cognitive Background Variables.

	JS	RD	LP	KI	FA	NK	Mean (*SD*)
Age (years)	11.3	11.7	12.9	13.3	13.5	13.6	12.7 (0.9)
Sex	M	F	F	F	F	M	
Level of lesion	T	L [a]	L	L	L	L	
Hydrocephalus and shunt (number of shunt revisions)	+ (3)		+ (0)		+ (0)	+ (0)	
Arnold-Chiari II	+	+		+	+	+	
Ventricles dilated	+		+	+	+	+	
Gait status							
Independent		+	+	+	+		
Partial						+	
Unable	+						
Handedness	R	L	R	R	R	L	
WISC-III means							
Verbal	9.8	11	9	9.5	11.8	13.5	10.8 (1.7)
Nonverbal	8.3	10.3	7.5	9.8	8.3	4.8	8.1 (2.0)*
Total[b]	9	10.6	8.3	9.7	10	9.1	9.4 (0.8)*

Notes. T = thoracic SBM; L = lumbosacral SBM; R = right; L = left.

[a]The hernia was partly occulta at birth.

[b]Since the WISC-III was administered in a short form (corresponding Donders, 1997), it was not possible to calculate intelligence quotients. The number for each individual refers to the mean scale score.

*$p < 0.05$, 1one-tailed Mann-Whitney test.

subtests to be appropriate for IQ-screening purposes. A verbal scale score mean was calculated from Similarities, Vocabulary, Arithmetic, and Digit Span. A nonverbal mean score was calculated on the basis of Picture Completion, Block Design, Coding, and Symbol Search, bearing in mind the limitations of the SBM group related to visuospatial and fine-motor skills previously discussed. The verbal and nonverbal means were averaged to form a total-scale-score mean. According to standardized WISC-III norms, the average range of scale scores run from 7 to 13, both scores included. The mean verbal score of the patients was 10.8 ($SD = 1.7$), which did not differ significantly from the control mean of 12.5 ($SD = 2.3$; $U = 28$, $p = .09$, one-tailed Mann Whitney). However, the nonverbal mean of the patients was only 8.1 ($SD = 2.0$), which was significantly lower than the control mean of 11.2 ($SD = 2.3$; $U = 14.5$, $p = .006$, one-tailed Mann Whitney). This was to be expected from previous studies, which generally show that children with SBM are at a disadvantage in nonverbal tasks that require quick manual skills, such as the ones in the WISC-III (Yeates et al., 2008). Mainly because of their low nonverbal scores, the total mean score of patients ($M = 9.4$, $SD = 0.8$) was also significantly lower than the control group's ($M = 11.9$, $SD = 2.0$; $U = 11.5$, $p = .003$, one-tailed Mann Whitney). However, it should be noted that all patients performed within the average range on both the verbal and the total scale score, and only one patient (NK) performed below average levels on the nonverbal scale. The children with SBM in this study can therefore be described as having normal intelligence.

Experimental Procedure

Experiments were run in E-prime (version 2.0). Subjects were placed in a dimly lit room in front of a 19" computer screen with a refresh rate of 100 Hz. Distance to the screen was held constant at approximately 70 cm. No head fixation or eye tracking was used, but subjects were reminded to stay still during trials and were offered the opportunity to move in between. Cerebellar damage may compromise the ability to perform smooth horizontal eye movements (Leigh & Ramat, 1999), but Salman, Sharpe, Lillakas, Dennis, and Steinbach (2009) found fixation of the gaze to be nonimpaired in SBM patients. Thus, fixation of the eyes was not considered to be more of a challenge to patients than controls.

During practice trials, eye movements were observed by the experimenter, and instructions to maintain fixation was motivated and repeated throughout the session (e.g. *"Keeping your eyes fixated at the center of the screen will make it easier for you to see the letters"*). Test instructions were given prior to practice and repeated if needed (between trials, not during testing). The experimenter was present during the full length of the tests to ensure the procedure ran smoothly, to clarify misunderstandings, and to motivate subjects. Subjects were tested for a time period of approximately 2 hours. The order of experimental paradigms was fixed, but the experimenter made sure that individual needs for breaks were met. All subjects were rewarded for participation with a gift voucher of DKK 250.

CombiTVA. The attentional parameters defined in the TVA model (C, K, t_0, α, w) were measured in a letter-identification experiment with two main conditions: whole and partial report. The former three parameters were measured using a whole report task (Duncan et al., 1999; Habekost & Starrfelt, 2009), where multiple visual elements are flashed at varying exposure durations. The task is simply to identify as many of the elements as possible. The number of correct reports increases systematically as a function of exposure duration and is analyzed by the TVA equations, the performance data yield estimates of t_0, C, and K (see Figure 1 for an example).

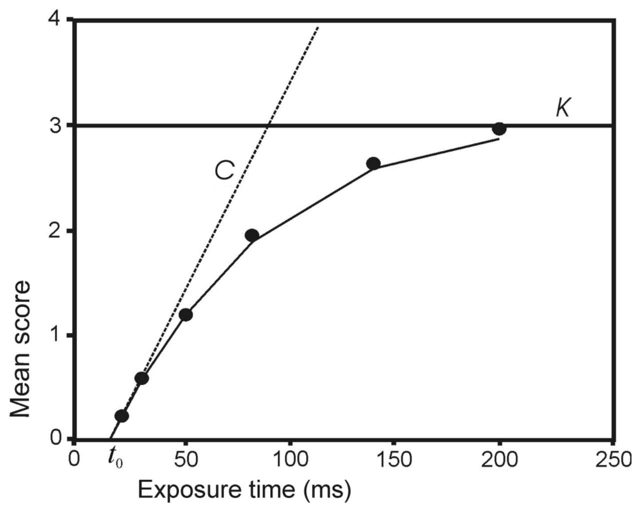

Figure 1 Whole report performance for a typical control participant in the present study. The panel shows the mean number of correctly reported letters as a function of exposure duration. Solid curves represent mathematical fits to the observations based on TVA analysis. The intercept with the x-axis corresponds to the perceptual threshold, t_0. The slope of the curve at the intercept with the x-axis equals the total visual processing speed for the stimuli in the display, C. The asymptotic level of performance corresponding to the maximum capacity of visual short-term memory, K, is marked by a horizontal line.

When distractor elements are added to the display (e.g., letters of another color than the target objects; partial report), the visual distractibility parameter, α, can be measured by comparing the accuracy reduction in this condition to trials without distractor elements. Finally, the relative attentional weight, *w*, of objects in the left versus right visual field can be estimated by analyzing performance when stimuli are shown simultaneously in both sides (Kyllingsbaek, 2006). The *CombiTVA* design used in this study was developed by Vankilde, Bundesen, and Coull (2011) and consists of nine test blocks of 36 trials (324 trials in total). Stimuli letters (A, B, D, E, F, G, H, J, K, L, M, N, O, P, R, S, T, V, X, and Z) were presented briefly on the screen for 20, 30, 50, 80, 140, or 200 ms, followed by a pattern mask (see Figure 2). Exposure durations and experimental conditions (whole or partial report) were varied systematically. Thus, exposure times span approximately from the threshold of visual perception (t_0) to the maximum exposure time in which eye movements are not possible (i.e., 200 ms). This choice of exposure times ensures the best measurement of the TVA parameters (Habekost & Starrfelt, 2009).

Target letters were red (RGB 255, 43, 43), distractor letters were blue (RGB 43, 53, 255), and masks consisted of a mixed pattern of the two colors. All stimuli were presented in font size 68. Reports were verbal and unspeeded. Participants were instructed to report as many red letters as possible, but to refrain from guessing. Feedback on error rates was given after each test block, and the participant was instructed to aim for a score of 80%–90% correct. Patients obtained a median score of 86.2% correct (range = 8.9), which did not differ significantly from the control median score of 86.7% (range = 12.7; $U = 44$, $p = .62$, two-tailed Mann-Whitney). Prior to testing, all participants completed two rounds

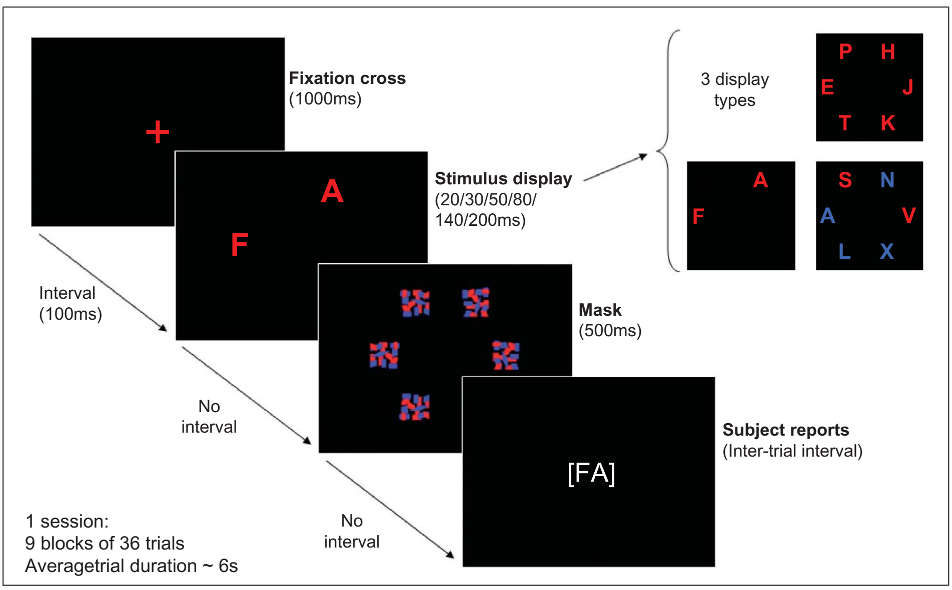

Figure 2 The CombiTVA test. The figure shows the outline of a single trial from the initial fixation to the verbal report (typed in by the experimenter). Also shown are the three different display types in the experiment. The participant must report as many of the red letters as possible, ignoring blue letters when these are present. [To view this figure in color, please visit the online version of this Journal.]

of practice (a total of 48 trials) to familiarize themselves with the task. Visual processing speed C was calculated for both sides of the display (C_{right} and C_{left}, respectively). Moreover, an index value was estimated on the basis of the formula $C_{\text{index}} = C_{\text{right}}/(C_{\text{right}} + C_{\text{left}})$. A C_{index} value of 0.5 indicates symmetric processing speeds in the two visual fields, whereas $C_{\text{index}} > 0.5$ indicates a higher processing speed of objects in the right visual field. Similarly, the tendency to attend left or right was calculated using the index formula: $w_{\text{index}} = w_{\text{right}}/(w_{\text{right}} + w_{\text{left}})$. Again, a w_{index} value of 0.5 represents a symmetric distribution of attentional weights in the two sides, but $w_{\text{index}} > 0.5$ indicates a distribution of attentional weights that favors the right display side.

For each participant the best-fitting set of TVA parameter values to the complete set of observed data was estimated. The fitting procedure was basically the same as in previous TVA based studies (Duncan et al., 1999; Kyllingsbaek, 2006) but was improved by a more robust fitting algorithm (Dyrholm et al., 2011). The new algorithm (a) does not assume that t_0 has a fixed value throughout the experiment but allows the parameter to vary from trial to trial following a Gaussian distribution and (b) does not assume that K varies between just two integer values, but that the parameter follows a wider distribution and equals a weighted average of values ranging from 1 to 6.

The Dual-Attention to Response Task (DART). In order to assess individual capacity for sustained attention and response inhibition, *the Dual-Task Attention to Response Task* (DART) developed by Dockree et al. (2006) was used. In DART, stimuli (numbers from 1–9) are presented in a fixed and predictable series above a centrally placed fixation cross. The exposure duration is 150 ms and stimuli are unmasked. In go trials, subjects are instructed to press the response key "1" following each white number except for the number 3 (no-go trials). In addition to this primary task, rare grey digits are presented without the predictability of the white digits. In response to these, subjects must press the response key "2" (see Figure 3). Measures are erroneous responses in no-go trials and to grey digits respectively, and reaction time is measured from stimuli offset. Responses made during stimuli exposure (from 0 to 150 ms after stimuli onset) are recorded as premature presses. Stimuli are presented in 25 test rounds, which allows for a progressive assessment of test behavior measured in both reaction time and error rates. DART takes approximately 7 minutes to complete. Digits are presented on a grey background (RGB 128, 128, 128); go stimuli were white (255, 255, 255) and rare targets grey (170, 170, 170). Stimuli size varied and digits were presented in font sizes 80, 90, 100, 110, and 120. Interstimulus intervals (ISI) varied between 1000 ms and 1500 ms to make the rhythm of presentation unpredictable. Different reaction time measures were analyzed in order to obtain detailed information on response behavior and to specify potential changes in responses throughout the test. Individual go-trial RTs were considered an indirect baseline measure of processing speed and error RTs as measures reflecting the RT expense of failed attention processes leading to an erroneous response.

Data Analysis

Patient performance was compared with control groups by means of two statistical methods. For comparisons between groups, the nonparametric Mann-Whitney test (SPSS version 18) was used, and statistical medians and ranges are reported. To assess error rates across trials in the DART task, repeated-measures analysis of variance (ANOVA) tests were used. Moreover, individual test scores were analyzed using a test devised by Crawford

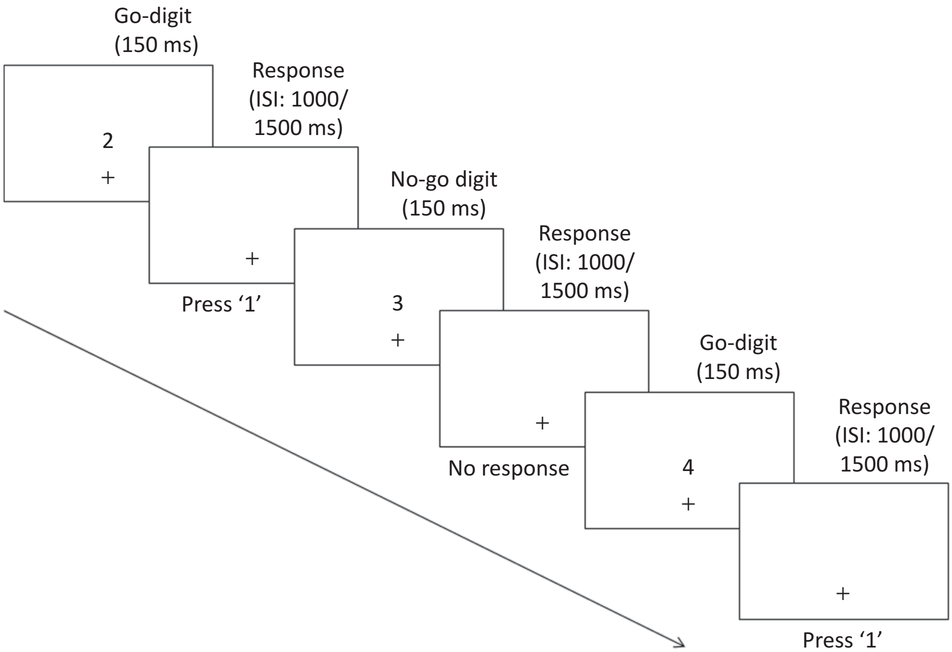

Figure 3 The DART test. An extract of the continuous sequence of stimuli is shown. Each digit is presented for 150 ms, followed by a response period of 1000 ms or 1500 ms. The participant must press "1" in go trials (all digits except 3) and withhold response in no-go trials (when 3 is shown). When unpredictable grey targets appear (not depicted here), the participant must press "2."

and Howell (1998). The test is based on the *t*-distribution instead of the usual normal distribution, which qualifies it to evaluate single-case results more robustly against means and standard deviations of control groups with limited size (Crawford & Gartwaite, 2002). In case of a control group with 18 participants (as in the current study), an individual score must deviate 1.8 standard deviations or more from the control mean to be judged significantly different (by a one-tailed test). The test has been employed in many single-case studies, including research based on the Theory of Visual Attention (Habekost & Rostrup, 2006). The *p* values reported are tested one-tailed in case of a theoretically based directional hypothesis (i.e., that patients would perform more poorly than controls), but otherwise two-tailed tests were employed.

RESULTS

CombiTVA

Group differences on TVA parameters are presented in Table 2. On a group level, only visual distractibility (the α parameter) was significantly elevated in children with SBM compared to controls ($U = 22.00$, $p = .044$; patient median = 0.82; control median = 0.42). Visual thresholds (t_0 values) in the patient group just failed to reach significance ($U = 24.00$, $p = .062$; patient median = 25.7 ms; control median = 17.2 ms). The patients'

Table 2 TVA Parameters.

	Group Data		Individual Data					
	SBM median (range)	Control median (range)	JS	RD	LP	KI	FA	NK
K	2.4 (1.6)	3.1 (2.9)	2.6	1.7* (−1.9)	2.3	3.2	2.3	3.3
t_0	25.7 (26)	17.2 (17.3)	18.3	27.3* (−1.8)	12.1	24.4	27.0* (−1.8)	38.2** (−4.0)
C_{right}	39.1 (36.4)	46.2 (61.5)	37.3	22.4	27.8	53.1	40.9	58.9
C_{left}	34.7 (46.9)	41.5 (40.6)	63.2	22.0	25.7	43.6	16.3* (−2.1)	56.1
C_{index}	0.52 (0.3)	0.49 (0.2)	0.37	0.5	0.52	0.55	0.72* (1.9)	0.51
α	0.82 (1)*	0.42 (0.7)	0.36	1.32** (3.9)	0.81	0.83	0.99* (2.3)	0.5
w_{index}	0.48 (0.3)	0.50 (0.3)	0.48	0.61	0.33	0.63	0.46	0.47

Note. K: capacity of visual short-term memory.; t_0: threshold for visual perception.; C_{right}: visual processing speed for objects in the right visual field.; C_{left}: visual processing speed for objects in the left visual field.; C_{index}: visual processing speed for objects in the right vs. left visual field.; α: the attentional weight of a distractor relative to a target (efficiency of selection).), w_{index}: the attentional weight of objects in the right vs. the left visual field. Apart from index values, which were tested 2two-tailed, all tests were 1one-tailed. For individual scores that deviate significantly from the mean of the control group, the magnitude of the deviation (in standard deviations) is given in brackets.

$^{*}p < 0.05$.

$^{**}p = 0.001$.

visual processing speed was generally within normal levels: In the right and left hemifield, the median values were $C_{right} = 39.1$ letters/s and $C_{left} = 34.7$ letters/s, respectively (compared to $C_{right} = 46.2$ and $C_{left} = 41.5$ in the control group: $U = 34.00$, $p = .256$ and $U = 35.00$, $p = .286$). Both patients and controls had approximately equal visual processing speeds in the right and left visual field, reflected in median C_{index} values of 0.52 of 0.49, respectively (no significant difference between groups: $U = 44.00$, $p = .624$, two-tailed Mann-Whitney test). Like the controls, patients also distributed attentional weights relatively symmetrically in space, without favoring either the right or the left display side (the median w_{index} in the patient group was 0.48; control median = 0.50; $U = 51.00$, $p =$ 1.0, two-tailed Mann-Whitney test). Although the median capacity of visual short-term memory, K, was only 2.4 in the patient group, compared to 3.1 in the control group, the patients' K values were not significantly reduced ($U = 29.00$, $p = .135$).

When compared individually, patients' results reveal a more nuanced picture (see Table 2). Patients JS, LP, and KI scored within normal limits of the control group on all parameters, while RD, FA, and NK deviated on one or more parameters. The threshold for visual perception (t_0) was significantly increased for RD (27.3 ms; $p = .042$), FA (27 ms; $p = .047$), and NK (38.2 ms; $p = .001$). RD also had a significantly reduced visual apprehension span (1.7 elements; $p = .035$). Visual processing speed (C) was significantly reduced in the left side of the display for FA (16.3 letters/s, $p = .024$) and because of a considerable difference between FA's left C value and the right-sided processing speed (40.9 letters/s), her C_{index} of 0.72 was significantly higher than controls, indicating a hemispheric asymmetry in visual processing speed resources ($p = .035$). Another patient, RD, had borderline C values, which failed to reach significance ($C_{right} = 22.4$ letters/s, $p = .053$; $C_{left} = 22$ letters/s, $p = .052$). Two patients (RD and FA) exhibited significantly elevated α values (RD: 1.32, $p = .001$; FA: 0.99, $p = .017$), and thereby seemed unable to filter out irrelevant information in this task. However, all patients performed within normal variability on the w_{index}, indicating a symmetric distribution of attentional weights.

DART

Response Errors. The numbers of commission and omission errors, grey stimuli detected, and premature presses were analyzed. Group medians are presented in Table 3. As a group, patients made significantly more errors of omission ($U = 21.00$, $p = .027$) and premature presses ($U = 24.5$, $p = .047$). The other two error measures in the patient group (commission errors and greys missed) did not differ significantly from the values of the controls.

Error scores from individual patients are also presented in Table 3. When compared on a case level, patients JS and RD performed within the range of normal performance on all error measures. Patients FA and NK made significantly more errors of commission than controls (FA: 13, $p = .011$; NK: 15, $p = .002$), and KI missed responding to a highly significant total of 28 times ($p < .001$), which corresponds to approximately 12% of all trials. Moreover, she responded prematurely with a significantly higher frequency than controls (14 times; $p < .001$), as did LP (7 times; $p = .004$).

Patterns of Reaction Time. Median group RTs are presented in Table 4. In terms of time to respond, children with SBM as a group did not on average score significantly different from controls on any RT measure (all RT measures were tested with two-tailed Mann-Whitney tests).

Table 3 DART Response Errors.

	Group Data		Individual Data					
	SBM median (range)	Control median (range)	JS	RD	LP	KI	FA	NK
Errors of commission (max. 25)	7.5 (15)	5.5 (8)	0	6	7	8	13* (2.5)	15* (3.2)
Errors of omission	8* (25)	4 (13)	3	8	8	28* (6.5)	9	7
Greys missed (max. 10)	5 (8)	6 (10)	9	7	4	1	6	2
Premature presses	3* (14)	1 (7)	0	3	7* (3.0)	14* (6.8)	3	3

Note. For individual scores that deviate significantly from the mean of the control group, the magnitude of the deviation (in standard deviations) is given in brackets.

*$p < 0.05$.

Table 4 DART Reaction Times.

	Group Data		Individual Data					
	SBM median (range)	Control median (range)	JS	RD	LP	KI	FA	NK
RT go trials	363.2 (501.9)	210.9 (211.9)	547.5*(5.4)	186.4	161.6	227.7	224.6	135.3
RT commission errors	205.5 (412.3)	280.2 (412.2)	–	363.2	579.4*(2.9)	660.3*(3.7)	334.2	158.4
RT greys detected	430 (538.7)	382.5 (336)	612.1	378.4	481.5	803.0*(3.8)	264.3	305.5

Note. All measures are presented in milliseconds. For individual scores that deviate significantly from the mean of the control group, the magnitude of the deviation (in standard deviations) is given in brackets.

* $p < 0.025$, 2two-tailed.

Individual RT scores, however, suggest a pattern of results more complicated than revealed by group analyses. Patient JS responded significantly slower on go trials (547.5 ms; $p < .001$, two-tailed Crawford and Garthwaite's test). Patients LP and KI, both of whom scored within the normal range on number of commission errors, spent significantly more time when they did make errors of commission (LP: 579.4 ms, $p = .011$; KI: 660.3 ms, $p = .002$, two-tailed Crawford and Garthwaite's test). This indicates a behavior opposite to the one exhibited by both FA and NK. These patients made significantly more errors of commission than controls but did so within the RT range of the controls. KI spent significantly more time to respond correctly to grey targets (803 ms; $p = .001$, two-tailed Crawford and Garthwaite's test). However, this result refers to only one correct response on the secondary task.

Group comparisons of the courses of significant error measures and go-trial RTs across Blocks 1–5 were investigated in repeated-measures analyses. For neither omission errors nor premature presses, the interaction effects of Block x Group were significant (as measured by a repeated-measures ANOVA test), indicating stable patient error rates across test blocks. Likewise, go-trial RTs did not decrease or increase significantly.

DISCUSSION

In this study we examined attention function in six children with SBM using a new set of experimental methods. The tasks employed did not have complex motor demands compromising data validity, were theoretically specific and should be sensitive to minor deficits. Besides introducing a new methodology for assessing attentional function in children with SBM, we particularly wanted to assess individual profiles of cognitive function to map the heterogeneity of the SBM syndrome. The methodology also made it possible to examine a number of attention parameters that have not previously been considered in the SBM literature. On a group level, the results showed poor attentional selection (α) for the children with SBM, consistent with predictions based on the existing literature. However, on the individual level, several patients did not have significantly reduced scores on this parameter. Three out of the six patients had significantly elevated perception thresholds (t_0), but this effect did not reach significance on a group level. Also, visual processing speed (C) did not generally differ from normal levels nor did the storage capacity of visual short-term memory (K); although there was a tendency for both parameters to be lower than in the control group. Visual perception was also generally symmetric. Sustained attention, however, was commonly affected in the patient group, as measured by several varieties of poor response inhibition (commission errors and premature presses) as well as frequent lapses of attention (omission errors). The evaluation of results on a case level provided detailed descriptions of the substantial individual variety displayed by the six patients: Despite the relative homogeneity of the group in terms of medical variables, the attention profiles of children with SBM were rather heterogeneous. The diversity of these profiles was not evident from group analyses.

The Patient Sample

When evaluating the results, it is important to keep in mind the specific characteristics of the patient sample. SBM is the most severe form of spina bifida and is more often than not associated with cerebral damage. Lesion level is a highly influential parameter determining the overall extent of neural damage, which makes lesion level important for cognitive development: low-level injuries are associated with better cognitive prognoses (Fletcher et al., 2005). As such, the patient sample may not be representative of the whole population of children with SBM: Even though all patients had open spinal injuries (as opposed to closed), JS was the only patient with a high-level injury. Furthermore, several factors of importance to cognitive development are important to notice in the patient sample. On the negative side, the presence of hydrocephalus, especially if severe enough to require shunting and revisions (as in the case of patients JS, LP, FA, and NK) is associated with more marked neurocognitive impairments compared to SBM with no hydrocephalus (Fletcher et al. 1995; Iddon, Morgan, & Sahakian, 1996). On the positive side, all patients except for JS had good gait status. The mobility of the individual patient is known to influence experience-based learning and is thus another parameter determining cognitive development (Dennis, 2000). Moreover, all patients had intellectual skills within the normal range: IQ served as criteria of inclusion to prevent confounding factors related to mental retardation. Considering the great variability produced by the level of the spinal lesion, the description of attention function provided by this study may apply most directly to SBM children with low-level dysraphisms. Although our group of patients may be classified as moderately affected compared to other, more severely disabling forms of SBM (high-level open spinal dysraphisms with mental retardation), we did find significant impairments of one or more attention functions in all six children. This may suggest a relatively high frequency of attentional deficits within SBM patients in general.

Basic Efficiency of Visual Attention Processes

One result reached significance on group level in the TVA results: impairments in attentional selectivity. Two patients (RD and FA) had significantly higher α values than normal, indicating poor ability to filter out irrelevant information. Another two patients showed nonsignificant borderline α values, 1.5 standard deviations above the control mean (LP: $\alpha = .81$ and KI: $\alpha = .83$). These results may help to explain the prevalence of the inattentive subtype of ADHD described in the SBM literature (Ammerman et al., 1998; Burmeister et al., 2005; Fletcher et al., 2005; Gadow & Sprafkin, 1987; Swartwout et al., 2008) and may correspond with the clinical impression of poor concentration and prevalent distractibility often observed even in well-adjusted SBM patients (Anderson, Northam, Hendy, & Wrennall, 2001; Wills, 1993). It seems plausible that if children are not effective at selecting relevant stimuli, processing becomes troublesome and time consuming. On a behavioral level, patients would therefore likely appear distracted and ineffective, simply because attentional resources necessary to focus on the relevant task are wasted on distracting events.

Although not significant on a group level, half of the patients (RD, FA, and NK) showed significantly elevated thresholds for visual perception (t_0). The impact of t_0 on everyday behavior must be interpreted with care. The median time at which children in the control sample started processing the visual display was 17.2 ms, and, compared to this, FA and RD were disadvantaged by 10 ms and NK by 21 ms. It is doubtful whether delays of this magnitude influence higher level mental processes like working memory or other complex functions. In other words, slightly elevated thresholds for perception may be compensated for during most everyday tasks where stimuli are available for inspection for more than 200 ms. Nonetheless, the results are interesting as they point to subtle deficits of basic visual processes in children with SBM, comparable in processing level to the orienting deficits described by Dennis et al. (2005a, 2005b). It is possible that some everyday tasks represent peak load situations in which even discrete perceptual impairments influence performance: Clinically, children with SBM often appear poorly coordinated in visually guided, goal-directed movements, and this was supported experimentally by Dennis, Fletcher, Rogers, Hetherington, and Francis (2002) who found action-based visual perception to be impaired in children with SBM as opposed to object-based perception. If processing onset is delayed corresponding to elevated t_0 values, visual information important to goal-directed movements might be lost, resulting in a coordination failure. For instance, successfully catching a flying football requires perception of a rapid sequence of movements, analyzed according to egocentric space in order to correctly predict the coordinates of the ball's descent and to initiate an appropriate movement of arms and hands.

Although reduced processing speed, based on reaction-time measures, has previously been reported by Fletcher et al. (1996) and Jacobs et al. (2001), we did not find that visual processing speed was significantly reduced in our patient group. The normal performance was found even though the CombiTVA includes near-threshold exposure durations, which puts high demands on visual processing speed. Only one patient (FA) met the criteria for significance, and only in the left visual field. The C values of patient RD were somewhat reduced compared to control means: RD's processing speed resources were borderline and close to significantly reduced in both visual fields, corresponding to 1.7 standard deviation below control mean. Although not significant, this may indicate the possibility of SBM-related reductions in processing speed resources in some but far from all children with SBM. The lack of significant reductions of processing speed in this study as opposed to the one by Jacobs et al. may be related to the experimental methods applied. Jacobs et al. used tasks that require the coordinated activity of complex motor function and visuospatial analysis (the Coding subtest of the WISC-III and The Rey-Osterrieth Complex Figure Test). Dependent variables in those tasks are time of completion, which as previously discussed may complicate interpretation and conclusions regarding central parameters of attention, for example, processing speed. In contrast, the verbal and unspeeded reports of the CombiTVA provide specific estimates of attention parameters on a perceptual level, not confounded by motor requirements. Therefore, it is possible that the significantly longer RTs found by Jacobs et al. can be ascribed to factors influencing behavior after basic perception has taken place, for example, factors related to motor timing and coordination. However, the question of visual processing speed in individuals with SBM needs further examination, and it seems crucial to take individual variability into account.

Only one patient, RD, exhibited reduced capacity of visual short-term memory, and was typically able to perceive only one or two objects at a time ($K = 1.7$). The cause for this reduction is unclear and somewhat surprising in view of RD's good status on medical variables compared to the other patients. Her lumbosacral hernia was partly occulta at birth, which should give her the advantage of less disabling neural damage (Gregerson, 1997). In accordance with this, she does not suffer from hydrocephalus and walks independently. Short-term memory in children with SBM remains to be described specifically in the SBM literature: Research up to now has been centered on the complex processes of explicit memory and recall. The detection of a significantly reduced capacity of short-term memory in a single, clinically well-functioning SBM patient establishes the heterogeneity of the neurocognitive profile of SBM and reflects the importance of highly sensitive case-based methods for both research and assessment.

Despite individual, nonsignificant variability, the group of patients distributed attention resources evenly in the two sides of the visual field. These results are not concordant with Dennis et al. (2005c), who found an exaggerated attentional bias to the left hemispace among 97 children and adolescents with SBM. Results were obtained using line bisection tasks, which also revealed a bias to inferior hemispace in this group. The line bisection task involves a motor component (marking the middle of a line with a pencil), and, although tasks were unspeeded, motor requirements could have influenced the results. The fact that the findings concerning spatial distribution of attentional resources are not concordant further supports the dissociation between visually guided movements and visual perception independent of motor output.

Capacity for Sustained Attention and Response Inhibition

The overall results from the DART showed that children with SBM made more hasty and poorly inhibited responses (as measured by premature presses). Moreover, the patient group experienced more lapses of attention than controls (as measured by omission errors); although only one patient's error rate was high enough to deviate significantly on an individual level (KI). Contrary to previous studies (e.g., Swartwout et al., 2008), we did not find significantly more commission errors in the patient group as a whole, only in two individual patients (FA and NK); a finding that may be attributed to lack of statistical power in our study.

The general results obscure important individual variations, which can be more elaborately understood at the case level by integrating the analysis of each patient's error and RT measures. In terms of response errors, patients JS and RD did not differ significantly from controls on any of the measures. However, to JS, this apparently normal performance had significant costs in RT on go trials: Compared to controls, he needed much more time to respond correctly. Patients LP and KI made more premature responses than normal, and in addition to this, KI demonstrated frequent attentional lapses. Both patients also spent significantly more time settling on a response when making commission errors, perhaps indicating an abnormally time-consuming process of inhibition (which eventually failed). It is likely that these results reflect different strategies for solving the DART. The performances of JS, LP, and KI may represent a behavior in which responses were held back and slowed in order to compensate for poor response inhibition. Both LP and especially KI, however, failed to be consequent in compensating and often responded in anticipation of the next target before it had appeared (premature presses). The performance of KI was further impaired by very frequent attentional absences (omission errors), which together with many hasty responses indicate an uneven, inattentive performance throughout the test. Perhaps the most extensive deficits of sustained attention parameters were found in patients FA and NK. Both of them made significantly more errors related to poor response inhibition (commission errors) but did not seem to be aware of this behavior: Their RTs were not prolonged, indicating no special attempt to compensate by carefully holding responses back until stimuli have been correctly perceived. On the contrary, NK in fact responded more quickly than controls in go trials, however not significantly faster.

In all, individual as well as group performances in the patient sample displayed quite pronounced SBM-related deficits in sustained attention, supporting the conclusions made by Brewer et al. (2001), Lollar (1990), Loss et al. (1998), and Swartwout et al. (2008). Like Swartwout et al., who showed consistent performance in children with SBM across test blocks in terms of go-trial RT, we found no deterioration of significant error rates (errors of omission and premature presses) or go-trial RT over the course of the testing session. This may indicate a stable, though perhaps tonically lowered, level of alertness, which makes it challenging to keep up appropriate response behavior in the absence of external reinforcement from salient stimuli. The individual severity of the deficit seems to vary considerably, which may lead to different behavioral strategies, all unsuitable to meet task demands: Responses made on the basis of too little information or excessively delayed responses. Both strategies are likely to affect the everyday life of children with SBM, causing the child to miss information important to learning and development.

Evaluating the Research Methods

The present study highlights the relevance of analyses at the individual case level. Although results were significant on a group level for parameters α, errors of omission, and premature presses, case evaluations made clear that no measures were significantly deviating for all of the six patients at a time. On parameters such as K, the number of commission errors, and reaction time for commission errors, the results reflect important variability hidden in the group data. Analyses on a case level thus allow for a more sensitive evaluation of the diversity of attention profiles in children with SBM. By taking individual variability into account, the present study may provide more specific information on attention function in SBM patients compared to previous descriptions in the literature.

As described in the introduction, the qualities of TVA whole and partial report paradigms have been solidly documented in many studies of adult neurological patients. The TVA-based test method has been able to identify specific yet subtle deficits, which were not detected in conventional tests. Moreover, the parameters of visual attention are very specifically described in a mathematical model that makes it possible to isolate measures that are otherwise entangled with other parameters. The present study confirmed the specific quality of TVA-based testing: Individually significant deficits on the t_0 parameter were dissociable from nonsignificant processing speed measures (C) and filtering mechanisms were found to be significantly affected on a perceptual level in two patients. Furthermore, this study pioneers the use of TVA for studies of children with neurological disorders. The simple instructions and task demands of the CombiTVA proved to constitute a well-suited tool for testing both normal and patient children aged 11–13. Lastly, the use of TVA in this study adds to the SBM literature by providing new information of attention function in children with SBM, independent of variables related to movement.

The CPT task DART, which is also new in the context of SBM research, used a fixed presentation sequence, which together with the second, distracting task should enable more sensitive measures of sustained attention and alertness. Alertness is not maintained by external factors of the experimental design (unpredictable stimuli), and as a consequence, the child's ability to keep up the appropriate level of alertness required to respond correctly and flexibly throughout the test should be the only variable affecting performance. Moreover, the additional RT measures available from the DART made it possible to nuance individual profiles of sustained attention. Go-trial RTs confirmed results presented by Dennis et al. (2005a) and Vinck et al. (2010) concluding that children with SBM are not disadvantaged by simple motor requirements (press of response keys). As a consequence, significant commission error RTs may be interpreted as reflecting specific deficits of response inhibition, an essential part of attention function. This enables descriptions of individual attention profiles of children with SBM that offer a clinically useful perspective on degrees of difficulty in addition to mere quantitative error rates.

The present study enhances understanding of the SBM condition and provides important methodological perspectives. The new methods and research hypotheses presented must be followed up by investigations of larger groups of patients that are more representative of the whole population of SBM patients. Specifically, it will be of great importance to discriminate between the influences of medical variables such as level of spinal injury. Such detailed descriptions of attention function in children with SBM may contribute to clinical practice by targeting interventions at key deficits, thereby increasing intervention quality.

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