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REGULAR ARTICLE

Learning, memory and executive functions in children with hydrocephalus

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Abstract

Aims: To explore learning, memory and executive abilities in children with hydrocephalus without learning disabilities, and to find out whether children with an isolated hydrocephalus differed from those with hydrocephalus in combination with myelomeningocele (MMC).

Methods: Thirty-six children with an intelligence quotient (IQ) of ≥70 from a population of all the 107 children with hydrocephalus born in western Sweden in 1989–1993 were examined and compared with age- and gender-matched controls. The neuropsychological assessment of the school-aged child (NIMES) test battery was used.

Results: The children with hydrocephalus differed significantly from controls in all functions apart from registration skills and recognition. Learning, memory and executive functions were all impaired. Twenty children with infantile hydrocephalus did not differ from those with hydrocephalus associated with MMC. Also, children with an IQ of >84 performed significantly worse than controls.

Conclusions: Despite an IQ of ≥70, children with hydrocephalus had significantly impaired learning, memory and executive functions. When major brain lesions resulting in learning disability had been excluded, the hydrocephalus, rather than the underlying aetiology, was most important for the development of cognitive functions.

INTRODUCTION

Hydrocephalus in children is a condition characterized by the enlargement of the cerebral ventricles due to increases in the pressure and volume of the cerebrospinal fluid in the ventricular system. It was found to have a prevalence of 0.8 per 1000 live births in the population-based study from which the children in this study were recruited (1). The aetiology is either prenatal and then, mainly due to malformations or intrauterine infections, with myelomeningocele (MMC) as the single most common cause, perinatal, most often caused by an intraventricular haemorrhage in children born preterm, or, more rarely, postnatal, with meningitis as the most frequent aetiology (1). The subgroup with MMC has a neural tube defect resulting from failure of the spinal cord to close properly during the first weeks of embryological development. More than 80% of these children also have a type II Arnold–Chiari malformation, a deformity of the brain stem and cerebellum that blocks the circulation of the cerebrospinal fluid, thereby causing hydrocephalus. Earlier studies of MMC focussed mainly on the physical consequences of the spinal lesion, such as the impairments in the extremities and bladder and bowel functions, the physical phenotype. The congenital brain abnormalities found in the cerebellum, midbrain and often in the corpus callosum, the neural phenotype, also result in cognitive impairments, the cognitive phenotype (2,3). This cognitive phenotype has attracted increasing interest in recent years (3–5).

Regardless of aetiology, the increased intracranial pressure causing hydrocephalus may result in secondary brain insults by stretching axons and compressing the white and grey matter, including cortical neurons, which may lead to cognitive and behavioural malfunction (6). About one-third of children with hydrocephalus have been reported to have an intelligence quotient (IQ) of <70 and another third to have an IQ in the low normal range of 70–85 (7). Irrespective of IQ level, there is a characteristic pattern of verbal intelligence being better preserved than nonverbal intelligence (7–9). Despite the preserved verbal intelligence, as is measured with traditional IQ tests, children with MMC and hydrocephalus have been shown to have pragmatic language deficits and poor reading comprehension (5,10–14). It has also been shown that children with congenital hydrocephalus have difficulties in learning and memory, problems that are not explained by low IQ scores alone. Mary Ann Scott et al. (15) found that, irrespective of aetiology, the children with hydrocephalus had a pervasive disturbance of memory processes, while children with hydrocephalus in combination with MMC have been shown to have a shorter memory span than controls (16), as well as a poorer explicit memory, defined as the conscious recollection of past events and experiences (17).

Executive functions are capacities that enable a person to engage successfully in independent, purposive, self-serving behaviour (18). This implies strategic thinking and planning capacities and the ability to shift attention, as well as self-monitoring and the ability to use feedback. Children with hydrocephalus often have impaired executive functions and deficits in problem-solving skills and strategic thinking (4,16,19,20).

The aim of this study was to explore the extent to which learning and memory, as well as executive functions, are impaired in children with hydrocephalus without learning disabilities (IQ > 69), and to find out whether children with an isolated hydrocephalus differed from those with hydrocephalus in combination with MMC and to compare their results with age- and gender-matched controls.

METHODS

Participants

Of all the 107 children born in 1989–1993 in western Sweden who were treated for hydrocephalus during their first year of life, 73 agreed to participate in a study of the cognitive outcome (7). One-third were found to have a normal intelligence (IQ 85–115), another 30% had an IQ of 70–84 and 37% had learning disabilities with an IQ of less than 70. Forty-seven children with an IQ of at least 70 were asked to participate in an elaborate study of learning, memory and executive functions. Thirty-six agreed (77%), 23 boys and 13 girls, aged 8–13 years, with a median IQ of 84 (range 70–112). Sixteen children had hydrocephalus in combination with MMC (median IQ 78, range 71–109; verbal IQ 88; performance IQ 76) and 20 children had an isolated hydrocephalus, referred to here as infantile hydrocephalus (median IQ 89, range 70–112; verbal IQ 94; performance IQ 82). Six children with infantile hydrocephalus had a posthaemorrhagic hydrocephalus and 4 children had a postinfectious hydrocephalus. In 6, the hydrocephalus was caused by a malformation and 1 child had a stenosis of the aqueduct. In 3 children, the aetiology was unknown. A revision of the shunt system had been performed in 22 children (61%), once or twice in 14 (39%), three or four times in 14 (22%) and more than four times in 2 (5%). These rates of revision were about the same as in the whole population of 107 children. Eleven of 47 (23%) children who were unwilling to participate did not differ from the study group in terms of gender (50% boys vs. 60% boys), aetiology (40% in both groups had MMC) or IQ (84 vs. 93; $p = 0.17$)

Thirty-six children in the study groups were compared to an age- and gender-matched control group of 36 healthy children from mainstream schools in the same region as the study groups. They were not formally assessed with IQ tests, but were considered to have a normal intelligence. The median age in both groups was 11 years and 7 months (range 8–13). Ten children (28%) in the study group and 4 children (11%) in the control group were left-handed ($p = 0.07$).

Assessments of the children in the study groups were performed by 9 psychologists at the child's local habilitation centre and in the control group by the first author (BL), who also assessed 11 of the children with hydrocephalus.

Instruments

The instrument that was used was a Swedish translation of a neuropsychological test battery for children 7–14 years of age, called 'neuropsychological assessment of the school-aged child' (NIMES), comprising 10 established and widely used neuropsychological tests of learning, memory and executive functions (Table 1) in the auditory-verbal and visuospatial domains. This test battery was standardized in 1997 in Australia (21) and in 2000 in Sweden (22), revealing a high concordance between the Australian and the Swedish findings. The Australian standardization is, therefore, recommended in the test manual to be also used for Swedish children.

Table 1 Tests in the neuropsychological assessment of the school-aged child (NIMES) measuring auditory-verbal and visuospatial functions in the form of registration skills, short- and long-term memory, learning and executive abilities

Function	Auditory-verbal	Visuospatial
Registration skills	Digit Span	Block Span
Short-term memory	Story Recall	Complex Figure of Rey Recall
Learning	Rey Auditory-Verbal Learning Test	Spatial Learning Test
Long-term memory	Story Recall	Complex Figure of Rey
	Delayed recall	Delayed recall
	Rey Auditory-Verbal Learning Test	Spatial learning
	Delayed recall	Delayed recall
Executive functions		Tower of London
	Verbal Fluency Test	Trail-Making test
		Complex Figure of Rey Organization

Registration skills

The Corsi block span (23) was used to test immediate memory for visuospatial material, where the child was asked to tap blocks presented by the examiner in sequences of increasing lengths. The Digit span (24) was used to assess immediate memory for serial auditory-verbal material. The children were asked to repeat as many digits as possible forward in the correct order. The tests measure the attention span (registration skill) for visual and verbal materials, respectively.

Learning and memory

Verbal learning and memory were assessed with Story Recall (25,26), where the children were asked to listen to two short stories and recall the content as correctly as possible. Additionally, they were examined with the Rey auditory-verbal learning test (RAVLT) (27,28), which consists of 15 words, where the children have five trials to learn as many words as possible read by the examiner. Scores were calculated for the total number of words recalled over the five trials, after inference, and the number of words recognized from the word list in a short story (recognition).

The Spatial Learning Test (26,29) is thought to assess the ability to learn and remember visuospatial material. The children are asked to memorize and recall the positions of nine pictures on a wooden board. The number of trials needed to fulfil the criterion was recorded, as well as spontaneous recall and recognition. The Complex Figure of Rey (ROCF) (30) is a task designed to assess visual memory, as well as visuoconstructional ability and visual organizing ability, where the children are asked to draw a copy of a complex figure with many details.

Long-term memory
After 30 min, the children were asked to recall information from the previous tasks: the stories (Story Recall), the 15 words (RAVLT), the positions of the pictures (without seeing the pictures) (Spatial Learning Test) and the complex figure (ROCF).

Executive functions
The ability for visual planning, strategy and mental flexibility was measured with the Trail-Making Tests A and B (28), where the children were asked to connect numbers and letters with a pencil as quickly as possible and to plan and organize the elements in the drawing of the ROCF. The problem-solving aspects of the executive functions were measured with the Tower of London Test (31). The children were given three wooden balls arranged on sticks and were asked to change the position of the balls in a similar manner to models presented on 12 cards in a prescribed number of moves. All tests of visual executive functions were time-limited. Executive aspects of language were measured with the Verbal Fluency Test (32), where the children were encouraged to generate as many words as possible in 1 min, beginning with F, A and S, respectively.

Recognition
Two tests of recognition (of spatial and verbal material) were also administered. Recognition abilities are not thought to reflect short-term memory, long-term memory or executive functions. The recall of spatial and auditive-verbal material by seeing or hearing a clue does not require strategic abilities to organize material to be stored in or retrieved from long-term memory.

Statistical analyses
All raw scores were converted to standard scores according to the Australian standardization (21), i.e. T-scores, where 50 is the mean (standard deviation [SD] 10). T-scores of ≤ 20 (i.e. ≤ -3 SD) were set at 20.

All T-scores were subjected to a univariate analysis of variance (ANOVA) based on the design: 3 groups $\times$ 15 tasks representing 5 functions (registration skills, short-term memory, learning, long-term memory and the executive functions of problem-solving and planning and organization) and 2 domains (auditive-verbal and visuospatial). The five functions and the two domains comprise mean composite T-scores for the respective test results. Significant group differences with more than two means were followed up with Tukey's Honestly Significantly Different (HSD) for unequal N (Spjotvoll–Stoline). Statistical analyses were performed using a PC-based statistical package (Statistica 7; StatSoft, Tulsa, OK, USA)

Ethics
The study was approved by the research ethics committee at Göteborg University. Informed consent was given by all the children and their parents.

RESULTS
The results for the three groups, infantile hydrocephalus, MMC with hydrocephalus and controls, on the tests of neuropsychological functions and their constituting subtests are presented in Table 2.

Registration skills
No significant differences were found between the two groups of children with hydrocephalus or between either of these groups and the controls regarding the subtests of registration skills. All three groups slightly underachieved in registration skills compared to the standardization, but the immediate recall of both visuospatial (Corsi block test) and auditory-verbal (Digit span) materials was fairly normal.

Short-term memory
A main group effect was found for short-term memory ($p < 0.001$). However, there was no difference between children with hydrocephalus with and without MMC. Both clinical groups obtained significantly lower results for short-term memory (about 1.5 SD under test norm) for both visuospatial and auditory-verbal materials compared with normal results in the control group.

Learning
The learning of spatial material ($p < 0.001$) and learning a word list ($p < 0.05$) was impaired in both clinical groups as compared to controls, but there were no significant differences in learning skills between the two groups with hydrocephalus. In spatial learning, the children with hydrocephalus needed more trials to fulfil the criterion (placing all nine pictures correctly on the board), or did not fulfil the criterion at all. The control group easily fulfilled the criterion and obtained results constantly 1 SD above the mean according to standardized norms.

Long-term memory
Both clinical groups were significantly inferior to controls on the two visuospatial long-term memory tasks ($p < 0.01$ and $p < 0.001$, respectively). For the children with MMC, the visuoconstructional task of drawing the Rey–Osterreich Complex Figure was shown to be more difficult than remembering the position of nine pictures in the spatial memory test. In both the story recall and the word list recall, the children with hydrocephalus performed 1.5 SD below controls and test norms. There were no significant differences between children with and without MMC.

Executive functions
Visuospatial planning and organization, measured with four subtests, revealed significantly greater problems for children with hydrocephalus who performed about 1 SD below norms than for controls ($p < 0.001$). Controls performed 1 SD over standard norms in the visuospatial planning tasks. In the Verbal Fluency Test, the clinical groups also achieved results significantly inferior to those of controls ($p < 0.001$).

The group differences shown in Table 2 were followed up with Tukey's HSD tests, which consistently revealed

Table 2 Means and standard deviations (SD) for five neuropsychological functions and the constituting subtests in NIMES for 36 children with hydrocephalus, 20 with infantile hydrocephalus (IH) and 16 with myelomeningocele (MMC) and 36 controls

Function and subtest	IH			MMC			Controls			p
	n	Mean	SD	n	Mean	SD	n	Mean	SD	
Registration skills	20	42.5	9.7	16	42.8	8.4	36	45.7	6.4	ns
Corsi block	20	44.1	11.3	16	45.1	9.7	36	47.9	10.4	ns
Digit span	20	40.8	10.5	16	40.5	10.1	36	43.4	8.2	ns
Short-term memory	19	36.5	11.1	14	33.3	9.7	36	52.2	9.2	***
ROCF recall	19	36.8	15.1	14	33.4	9.8	36	53.9	14.4	***
Story recall	20	36.2	13.4	16	34.5	13.3	36	50.5	8.0	***
Learning	20	40.7	16.2	16	40.8	9.2	36	54.8	7.3	***
Spatial learning	20	42.2	21.8	16	44.4	12.8	36	60.3	7.8	***
RAVLT 1–5	20	39.2	14.8	16	37.1	13.7	36	49.3	12.2	*
Long-term memory	20	37.5	12.8	13	35.3	8.2	36	51.2	8.0	***
Spatial delayed recall	20	39.6	16.3	16	40.6	13.1	36	49.6	9.2	**
ROCF delayed recall	20	36.5	14.4	13	32.8	12.0	36	53.1	15.1	***
Story delayed recall	20	32.6	13.7	16	31.1	9.0	36	47.2	7.6	***
RAVLT delayed recall	20	41.5	17.6	16	36.3	13.0	36	54.8	11.6	**
Executive functions	19	41.7	9.0	14	41.4	8.8	36	55.0	4.4	***
Trail-Making Test A	20	39.1	14.3	16	38.6	11.0	36	53.1	6.7	***
Trail-Making Test B	20	39.3	12.0	16	40.9	14.0	36	52.0	6.4	***
ROCF organization	20	47.8	16.4	14	41.4	13.1	36	60.4	13.3	***
Tower of London	19	38.4	10.9	16	40.1	14.4	36	58.6	5.9	***
Verbal fluency	20	42.8	12.6	16	45.2	9.6	36	50.5	12.0	***

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.
ROCF = Complex Figure of Rey; RAVLT = Rey Auditory-Verbal Learning Test.

significant differences between controls and the two clinical groups in all the subtests apart from Corsi blocks and Digit span (registration skills) ($p = 0.17$ and 0.21, respectively).

Accordingly, when comparing the two domains of auditory-verbal and visuospatial abilities, the children with hydrocephalus obtained significantly poorer results, about 1 SD below the mean from standardized norms (Table 3). There was no difference between the domains.

Recognition

There were no statistically significant differences in auditory-verbal recognition between children with hydrocephalus and controls, or between children with hydrocephalus with or without additional MMC. All three groups produced average results according to test norms. However, in the spatial recognition task, the children with hydrocephalus performed significantly below controls ($p < 0.01$).

Table 3 Means and standard deviations (SD) in auditory-verbal and visuospatial domains of neuropsychological functions in 36 children with hydrocephalus, 20 with infantile hydrocephalus (IH) and 16 with myelomeningocele (MMC) and 36 controls

Domain	IH			MMC			Controls			p
	n	Mean	SD	n	Mean	SD	n	Mean	SD	
Auditory-verbal	20	38.8	8.1	16	37.5	7.4	36	49.3	6.8	***
Visuospatial	18	40.4	10.1	13	39.6	7.0	36	54.3	5.0	***

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.

Effects of IQ

There were no differences between children with infantile hydrocephalus and children with MMC with respect to IQ. To estimate the impact of the IQ level in all children with hydrocephalus on their results, they were divided into two groups: one with an IQ over 84 (mean 98, $n = 17$) and one with an IQ of 70–84 (mean 77, $n = 19$). In the former group, the children with infantile hydrocephalus, as well as those with MMC, had a mean IQ of 98. The corresponding mean values in the lower IQ groups were 78 and 76, respectively.

The IQ groups did not differ in terms of learning and long-term memory, but the children with an IQ of >84 had significantly better results in short-term memory tasks ($p < 0.05$) and executive functions ($p < 0.001$), as well as in the auditory-verbal ($p < 0.05$) and visuospatial ($p < 0.001$) domains. As the control group was selected from mainstream schools, the children were assumed to have a normal intelligence. When comparing the children with hydrocephalus and a normal IQ with the controls, there were still significant differences for all functions except registration skills.

DISCUSSION

Despite a normal or near-normal IQ, the children with hydrocephalus were found to have impaired learning, memory and executive functions; only recognition and registration skills were normal. IQ, however, contributed strongly to the poor short-term memory and impaired executive functions. No difference was found between those with MMC and those with other aetiologies of the hydrocephalus.

The finding that children with hydrocephalus did not differ from controls in terms of recognition and registration skills may be explained by their preserved ability to register material that is not supposed to be stored and subsequently remembered. The immediate recall of numbers and imitating pointing at blocks do not require analysis (implicit memory). To memorize and recall a story, even if it is very short, on the other hand, requires this kind of analysis (explicit memory). That children with hydrocephalus (and MMC) did not differ from children without brain injuries in terms of implicit memory, but obtained significantly poorer results for explicit memory, was also found by Yeates et al. (17). This finding may be explained by the hypothesis that implicit memory is mediated by less specialized brain systems in children than in adults and is, therefore, less vulnerable (17).

Significant verbal and nonverbal memory deficits have been reported in children with MMC and infantile hydrocephalus (15–17,33). This study revealed that children with hydrocephalus had significant difficulties to recall short stories, both immediately and after 30 min.

Moreover, visuospatial memory in children with hydrocephalus differed significantly from that in controls, both short-term and long-term memory, especially the spatial memory task of the ROCF, which appeared to be much more difficult than the spatial memory task where the children were asked to memorize the position of pictures. The latter task was probably easier because of the familiarity of the test material, compared with the unknown ROCF, and because of the different demands imposed on the child, i.e. motor-free visual registration of pictures versus the more effortful visuoconstructional task of copying.

The recognition of verbal material was not impaired in children with hydrocephalus. This finding was corroborated by Yeates et al. (34). They still suggested that these children had word-retrieval deficits; although the information had entered long-term storage, the children had an impaired development of organizational strategies for effective storage and retrieval. However, if they were helped with a cue, they were able to recall the information they had learned as well as the controls.

The learning, retrieval and recall of verbal and spatial material, therefore, appear to be a core problem in children with hydrocephalus independent of aetiology. Vachha and Adams (16) showed that children with MMC had a narrower memory span than controls, as well as ineffective learning strategies, reflecting poor executive functions. When comparing children who had required shunt treatment for their hydrocephalus to children with spontaneously arrested or no hydrocephalus, Scott et al. (15) found that only children with shunted hydrocephalus had memory problems.

In the present study, the children with hydrocephalus had significantly impaired executive functions in the form of visual planning and strategic thinking. It could be argued that the poor performance on the Trail-Making Test and Tower of London Test in this study may reflect deficits in fine motor functions, which are well known in children with MMC, rather than executive dysfunctions. However, the children with hydrocephalus without MMC performed equally poorly, indicating that the results were instead due to planning and speed deficits.

It has recently been shown that children with MMC and hydrocephalus have relative strengths on visual perception tasks involving categorical relationships (object perception, face recognition and illusions of size and length), but relative deficits on figure ground, mental rotation and illusory figures and action-based perception (visual tracking, drawing and route finding) (5). Visual strategy problems were confirmed in this study by the results from copying the ROCF and the Tower of London Test, where the two groups of children with hydrocephalus had significant deficits in strategic ability.

The auditory-verbal planning task also revealed a difference between children with hydrocephalus and controls. Children with hydrocephalus generally have fluent language (10), but, in the present study, they had severe problems in performing the word-finding task.

The earlier well-documented discrepancy between verbal and visuoperceptual abilities in children with hydrocephalus (7–9) was not confirmed in this study when it came to the verbal and spatial domains. It has been shown that the relatively strong verbal abilities, as measured with intelligence tests are of limited value for predicting good language function. These tests include single word knowledge, which is very good in children with hydrocephalus, but does not measure abilities in pragmatics or discourse where these children have great difficulties (10–13). Correspondingly, children with hydrocephalus in this study displayed serious difficulties to learn and remember short stories compared to the word list, which may be a consequence of discourse problems.

Most research on children with hydrocephalus has focussed on the group with MMC, which is natural because of its homogeneity and also because it represents a relatively large group of congenital childhood impairments with lifelong dysfunctions in cognitive and motor abilities. In this study, we included all children with hydrocephalus from a population-based study with the inclusion criterion of an IQ of at least 70. The aetiology of infantile hydrocephalus in this study was heterogeneous, but the results from neuropsychological measures were homogeneous, suggesting that the consequences of hydrocephalus *per se*, with its influence on white and grey matter structures, overshadow the effects of the congenital brain malformations in children with MMC. For further understanding, it would be of interest to study neuropsychological functions in the small group (less than 20%) of children with MMC that do not develop hydrocephalus.

One could argue that the participation of nine psychologists would imply low test reliability, but the fact that the psychologists were experienced and that the children and parents did not have to travel far or to meet too many new persons should outweigh this risk.

In conclusion, children with hydrocephalus were found to have major deficits in learning, memory and executive functions. These findings emphasize the importance of neuropsychological examinations of children with hydrocephalus in order to understand their special needs in school and in society and to optimize participation and quality of life.

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