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Case study

Short- and long-term neurobehavioral effects of lumbar puncture and shunting in patients with malabsorptive hydrocephalus after subarachnoid haemorrhage: An explorative case study

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ABSTRACT

Background: The neuropsychological effects of lumbar puncture and shunting in terms of cognitive functioning and quality of life were analyzed prospectively in four patients with malabsorptive hydrocephalus who became symptomatic in the chronic state after aneurysmal subarachnoid haemorrhage (SAH).

Methods: A comprehensive battery of neuropsychological tests was applied to four patients before and shortly after lumbar puncture and six months later. In three of them a shunt has been inserted, one patient was treated by repeated lumbar punctures. In addition, the patients completed a quality of life and a depression questionnaire before lumbar puncture and after shunting. The data were analyzed using single-case methodology.

Results: Hydrocephalus was associated with pronounced cognitive deficits in terms of functions of attention, short- and long-term memory, concentration and motor fine-coordination but not with a general mental deterioration. Quality of life and affect were also substantially impaired. Neuropsychological tests of fronto-cortical cognitive capacity, motor fine coordination and reaction time proved to be sensitive for the short-term effects of lumbar puncture. Memory functions and the capacity of divided attention needed more time for regeneration and improved substantially after shunt implantation.

Conclusions: We found a complex pattern of cognitive improvement after lumbar puncture and shunting. Furthermore, our results also show a typical cluster of cognitive deficits associated with malabsorptive hydrocephalus including motor dysfunction. These preliminary findings should be confirmed in larger patient samples.

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1. Introduction

Malabsorptive hydrocephalus (MH) is a frequent late complication of subarachnoid haemorrhage (SAH) developing in 10–43% of the patients [1]. In patients with normal pressure hydrocephalus (NPH) the indication for shunting is dependent on the clinical improvement of the patient after lumbar puncture (LP) [2]. In contrast, little is known as of yet about the neuropsychological consequences of LP and shunting in patients with MH after SAH [3]. It is not clear which neuropsychological functions are particularly sensitive for changes of higher cerebral functions due to intracranial pressure. The present study was performed to explore the immedi-

ate cognitive changes after LP and the long-term course of cognitive functioning and quality of life in patients with MH after SAH.

2. Patients and methods

2.1. Patients and procedure

The inclusion criteria were as follows: the age at the time of haemorrhage was 18–65 years; increased intracranial pressure caused by acute MH in the postacute or chronic state not later than one year after aneurysmal SAH; no further neurological and/or psychiatric disease and any additional severe internal illness; a good neurological result (GOS = I) according to the Glasgow Outcome scale [4] at discharge; good German language competency. Informed consent could be obtained from five of seven patients fulfilling the required criteria during the study period. One additional patient refused further participation at the study. Severity of SAH

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was classified according Fisher [5] while the clinical state of the patients on admission was rated according to Hunt & Hess [6]. The first neuropsychological examination was performed immediately before LP, a second 30–45 min after the release of cerebrospinal fluid (CSF) and a follow-up about half a year later. In addition, quality of life and depression were assessed at the time of the first neuropsychological examination and at follow-up.

2.2. Neuropsychological tests used

Handedness was determined by the Edinburgh Interview [7]. Speed of motor fine-coordination in both hands was measured by the Finger Tapping test [8], concentration capacity by the test d2 [9]. The memory scale of the German intelligence test IST was used to examine verbal long-term memory [10]. The German version of the Recurring-Figures and the Recurring-Words test was employed for measuring figural and verbal short-term memory [11,12]. The Benton test served to examine visuoconstructive capacity [13]. Further, by means of three tasks of a computerized neuropsychological testing battery (TAP) [14] reaction time, alertness, divided attention and selectivity were assessed. In addition an alertness score was computed [14]. The examination of divided attention demands the simultaneous pursuit of two different tasks [14]. The Go/NoGo test is a task of selectivity, a fronto-cortical cognitive function [14]. Finally, we used the Mini-Mental State [15] exam. For the memory tasks parallel versions for the different examinations were used.

2.3. Analysis of neuropsychological test scores

In order to ascertain the individual level of cognitive capacity, the test scores were compared to the published age-adjusted test norms. A deficit is indicated if the test score reaches or underscores a value two standard deviations below the mean of the test norms. The Benton test has individual cut-off values to indicate a deficit [13]. However, because of a wide range of normal scoring in the Finger Tapping test, the computation of test norms and their application is not useful [8]. This measure is quite sensitive to detect intraindividual change [8] but has the drawback that critical difference scores cannot be computed as well as test norms [16,17].

2.4. Examination of quality of life

Quality of life was examined by the Aachen Life Quality Inventory (ALQI) [18]. The ALQI was developed for the use in patients with brain damage. It contains 117 items in 11 subscales, which enable the calculation of 11 subscale impairment scores, a sum score of total impairment (Total score), a psycho-social (Social score) and a physical summary score (Physical score). The ALQI consists of a self-rating and a parallel proxy-rating version.

2.5. Assessment of depression

Depression was measured by the Beck Depression Inventory (BDI) for self-administration and by the Hamilton Depression Rating scale [19,20]. In the BDI a raw score of >10 and in the Hamilton Depression Rating a raw score >9 was regarded as an indication for the presence of a depressive disorder.

2.6. Statistics – the single case methodology

In order to confirm statistically the differences between the test scores of each individual patient, the critical cut-off values were computed according to Huber [17]. All statistics needed for this single-case statistical procedure were taken from published test norms. All cut-off values gathered by this procedure represent crit-

ical differences significant at a type-1 error level of $p < 0.05$ with two-sided testing [16,17]. In single-case studies these procedures allow the statistical confirmation of differences found in parallel tests or in the case of repeated measurement [16,17].

3. Results

3.1. Case 1 patient K.A.

A 54 year-old right-handed female (Edinburgh = +93) had sustained SAH from a ruptured aneurysm of the right middle cerebral artery (MCA). On admission the patient was in a Hunt&Hess grade III. The preoperative cerebral computed tomography (CCT) showed a right-frontal intracerebral haemorrhage and a thin layer of intraventricular blood in both lateral ventricles (Fisher grade 4). The aneurysm was clipped two days after the bleeding. Two days after surgery, the patient developed a psycho-organic syndrome and vasospasm. Three weeks later, the patient was discharged home without any neurological or psychiatric disturbances. A CCT scan performed three months later revealed a right frontal hypodense lesion of 2×3 cm and no further abnormalities. Eight months later, she was readmitted to the neurosurgical department because of cognitive slowing, memory disturbances, disorientation, gait disturbances and urinary incontinence. The Mini-Mental State score was 23. In addition to the known right frontal substance defect, a CCT scan showed a distinct dilatation of the ventricle system, interpreted as a substantial MH. The CSF pressure was 27 cm H₂O and 35 ml liquor were drained. Neuropsychological examination was performed before and after LP. On the day after the insertion of a ventriculo-atrial shunt, the mental symptoms, gait disturbances and the urinary incontinence became rapidly less severe and resolved completely in the further course. Four months after shunt insertion when the neuropsychological follow-up examination was performed, magnetic resonance imaging (MRI) demonstrated the correct position of the shunt, a slight symmetrical enlargement of the lateral ventricles and modestly increased periventricular signal intensities. Table 1 gives the neuropsychological data of this patient.

3.2. Case 2 patient T.C.

A 53 year-old right-handed (Edinburgh = +100) male patient suffered SAH following the rupture of an anterior communicating artery aneurysm (ACoA). He was in a Hunt&Hess grade IV on admission. On CCT scan a frontobasal SAH with intraventricular blood in the posterior horns of both lateral ventricles was seen (Fisher grade 4). The postoperative course was uneventful. A mild psycho-organic syndrome resolved rapidly after surgery. Daily TCD investigations showed no signs of vasospasm. The patient was discharged six days after SAH to another hospital with no neurological deficits or dysfunctions. A CCT scan performed 4 weeks later gave evidence of a small frontal hygroma and slightly enlarged lateral ventricles but no parenchymal focal lesions. One year later, the patient was admitted to the neurosurgical unit because a CCT scan demonstrated a ventricular widening accompanied by cognitive slowing, memory problems, tiredness, gait disturbances and urinary incontinence. The Mini-Mental State examination revealed a score of 21. The CSF pressure was 30 cm H₂O and 15 ml CSF were drained. After implantation of a lumbo-peritoneal shunt, the clinical state improved rapidly. Four days after shunting, he could return home in a good clinical condition. Table 2 gives the neuropsychological test results of this patient. The follow-up exam was performed 28 weeks after shunt insertion.

Table 1
Neuropsychological performance of patient K.A. (case 1).

Neuropsychological test used	Cognitive function tested	Before lumbar puncture	45 min. after lumbar puncture	Sign. difference	Follow-up	Sign. diff. 1st assessment/follow-up
Finger Tapping test right	Motor fine coordination	21.00	25.50	a	41.25	a
Finger Tapping test left	Motor fine coordination	15.50	19.25	a	26.50	a
d2 concentration speed ¹	Concentration capacity	82 ⁻	85 ⁻	0	89 ⁻	+
d2 concentration errors ¹	Concentration accuracy	72 ⁻	82 ⁻	+	89 ⁻	+
Benton test ²	Figural short-term memory	2 ⁻	3 ⁻	0	6	+
Recurring Figures test ³	Figural short-term memory	20 ⁺	20 ⁺	0	39	+
Recurring Words test ³	Verbal short-term memory	20 ⁺	20 ⁺	0	39	+
IST Memory ¹	Verbal long-term memory	84 ⁺	84 ⁺	0	102	+
Simple reaction time (ms) ⁴	Cognitive speed	571 ⁺	450 ⁻	+	233	+
Signalled reaction time (ms) ⁴	Cognitive speed	464 ⁺	452 ⁻	0	189	+
Alertness task ³	Alertness	55	43	-	66	+
Divided attention omissions ³	Attention capacity	20 ⁻	20 ⁻	0	42	+
Divided attention reaction time ⁴	Attention capacity	638	1120 ⁺	-	737	-
Go/NoGo task errors ³	Selectivity	20 ⁻	20 ⁻	0	57	+
Go/NoGo task reaction time ⁴	Selectivity	586 ⁺	475	+	413	+

^a For statistical reasons no significance testing possible.

¹ Standard scores.

² No. of correct reproductions.

³ T-scores.

⁴ Median reaction time in milliseconds (ms). Specification of signs: – significantly worse ($p < 0.05$); + significantly better ($p < 0.05$); 0 no significant difference.

^{*} Marks a cognitive deficit.

Table 2
Neuropsychological performance of patient T.C. (case 2).

Neuropsychological test used	Before lumbar puncture	45 min. after lumbar puncture	Sign. difference	Follow-up	Sign. diff. 1st assessment/follow-up
Finger Tapping test right	21.25 ⁺	20.75 ⁺	a	22.25 ⁺	a
Finger Tapping test left	22.25	26.75	a	35.25	a
d2 concentration speed ¹	95	95	0	100	0
d2 concentration errors ¹	87 ⁺	100	+	100	+
Benton test ²	2 ⁻	3 ⁻	0	7	+
Recurring Figures test ³	28 ⁺	20 ⁺	0	39	+
Recurring Words test ³	25 ⁺	26 ⁻	0	28 ⁻	0
IST Memory ¹	84 ⁺	86 ⁺	0	80 ⁺	0
Simple reaction time (ms) ⁴	380 ⁺	241	+	221	+
Signalled reaction time (ms) ⁴	402 ⁺	256	+	249	+
Alertness task ³	42	26 ⁻	-	48	0
Divided attention omissions ³	34	26 ⁻	-	38	0
Divided attention reaction time ⁴	813 ⁺	621 ⁺	+	634 ⁺	+
Go/NoGo task errors ³	20 ⁻	41	+	45	+
Go/NoGo task reaction time ⁴	514	454	+	392	+

^a For statistical reasons no significance testing possible.

¹ Standard scores.

² No. of correct reproductions.

³ T-scores.

⁴ Median reaction time in milliseconds (ms). Specification of signs: – significantly worse ($p < 0.05$); + significantly better ($p < 0.05$); 0 no significant difference.

^{*} Marks a substantial cognitive deficit.

3.3. Case 3 patient M.F.

This 46 year-old right-handed male patient (Edinburgh = +87) sustained SAH from a large aneurysm of the tip of the basilar artery. On admission he was in Hunt&Hess grade III. A CCT scan showed blood in the basal cisterns and in both Sylvian fissures (Fisher grade 3). The aneurysm was occluded by coiling. Angiography performed immediately after the intervention revealed a thrombotic occlusion of the posterior main trunk of the right MCA. This thrombus was lysed immediately with the aid of locally applied rtPA, leading to a restitution of the blood flow. A CCT scan performed 15 days after SAH revealed a symmetric ventricular enlargement. The Mini-Mental State showed a score of 29. A lumbar puncture revealed a CSF pressure of 25 cm H₂O and 25 ml were drained. CSF was drained by LP every second day which led to a consistent improvement of the clinical state. Because intracranial pressure diminished continuously, no indication for shunting was seen. The patient was discharged neurologically intact and com-

pletely mobile. On CCT scan no parenchymal lesions, a stable position of the coil but a slurred relief of the cortex were seen. Table 3 presents the results of his neuropsychological examinations while the follow-up was performed 30 weeks after the acute treatment.

3.4. Case 4 patient G.R.

This 50 year old right-handed male patient (Edinburgh = +100) suffered SAH from a ruptured ACoA aneurysm. He was admitted in deep coma, vegetatively instable in a Hunt&Hess grade V presenting with extensor posturing in arms and legs on pain. A CCT scan demonstrated blood in all basal cisterns including the prepontine cistern and thick layers of blood covering the temporal cortex on both sides (Fisher grade 3). The patient was operated upon within 24 h and the aneurysm could be clipped. A CCT scan performed 4 days later proved the correct position of the clip, a demarcation of a 2 cm large bifrontal paramedian infarction and a swelling of the right temporal lobe. When the patient was

Table 3
Neuropsychological performance of patient M.F. (case 3).

Neuropsychological test used	Before lumbar puncture	45 min. after lumbar puncture	Sign. difference	Follow-up	Sign. diff. 1st assessment/follow-up
Finger Tapping test right	23.75	25.25	a	24.25	a
Finger Tapping test left	17.00	22.00	a	17.75	a
d2 concentration speed ¹	76 [*]	80 [*]	0	111	+
d2 concentration errors ¹	72 [*]	77 [*]	0	98	+
Benton test ²	5 [*]	6	0	8	+
Recurring Figures test ³	28 [*]	20 [*]	0	43	+
Recurring Words test ³	29 [*]	20 [*]	0	38	+
IST Memory ¹	84 [*]	86 [*]	0	100	+
Simple reaction time (ms) ⁴	255	287	0	202	+
Signalled reaction time (ms) ⁴	199	211	0	184	+
Alertness task ³	69	80	+	56	–
Divided attention omissions ³	20 [*]	20 [*]	0	34	+
Divided attention reaction time ⁴	972 [*]	743	+	872 [*]	0
Go/NoGo task errors ³	20 [*]	31	+	46	+
Go/NoGo task reaction time ⁴	664 [*]	494	+	372	+

^a For statistical reasons no significance testing possible.

¹ Standard scores.

² No. of correct reproductions.

³ T-scores.

⁴ Median reaction time in milliseconds (ms). Specification of signs: – significantly worse ($p < 0.05$); + significantly better ($p < 0.05$); 0 no significant difference.

^{*} Marks a cognitive deficit.

referred to another hospital he had no focal neurological deficits but presented with a psychoorganic syndrome. This resolved completely two months later. Five months later, he was readmitted to the neurosurgical department. Beyond the known bifrontal paramedian infarction a CCT scan revealed a substantial increase of the size of all ventricles indicative of MH. The patient presented with pronounced memory disturbances and mental slowing but was not aware of his problems. The Mini-Mental State score was 30. CSF pressure was 30 cm H₂O and 30 ml were drained. After insertion of a ventriculo-atrial shunt the patient improved rapidly and could be discharged home. The neuropsychological follow-up took place 36 weeks later. Table 4 gives the results of the neuropsychological examinations.

3.5. Quality of life

Table 5 presents the time course of the quality of life and depression.

4. Discussion

In contrast to earlier studies and case reports about the cognitive concomitants of hydrocephalus [21–24], our patients had MH of the same type and etiology. In every patient the Mini-Mental State score was >20 (21–30) and therefore, in spite of their substantial focal cognitive impairments, they were not demented. Similar findings of uncritical Mini-Mental State scores in patients with acute hydrocephalus were reported by Smyss et al. [26]. One of our patients had developed a MH in the postacute stage after SAH. While the other patients were treated by shunt insertion, this patient received repeated LP which proved to be effective in this case also in the long-term perspective.

4.1. Neuropsychological effects of LP and shunting

With acute hydrocephalus, marked deficits were prominent in motor fine-coordination, figural and verbal short- and long-term memory, reaction time, divided attention, selectivity and/or slowing of the reaction time in the Go/NoGo task. This pattern of cognitive impairments with predominant memory and attention deficits has also been described by other authors [3,23,24,27]. Immediately after LP the most consistent improvements occurred in all cases in

at least one parameter of the Go/NoGo task, which is particularly sensitive for fronto-cortical functional capacity [14]. This is in close correspondence with several PET studies which demonstrated in communicating, obstructive or so-called NPH the most pronounced disactivation in fronto-cortical areas around the third ventricle, the temporal horns of the lateral ventricles and in the basal part of the frontal lobes [28,29]. Moreover, Torkelson and coworkers [29] described that after lumbar puncture or after shunting the fronto-cortical disactivation disappeared in PET scans. A pronounced improvement in the speed of motor-fine coordination (Finger Tapping) was observed in all of our patients shortly after LP. This suggests that hydrocephalus is also associated with pronounced disturbances of motor fine-coordination. Therefore, the Finger Tapping task may be useful as a simple screening procedure for the effectiveness of LP and/or shunting on mental function although it is impossible to compute reliable test norms [8,16,17]. Clear and consistent improvements in simple reaction time and the accuracy of concentration were seen in three of our patients shortly after LP. Therefore, the measurement of simple reaction time could also be suitable for assessing changes in CSF dynamics. The observation, that shortly after LP a total of 35% (18/52) of the cognitive parameters improved substantially, while at follow-up 69% (27/39) improved, indicates that the process of restoration takes some time, particularly in memory functions. This is in line with the findings of some earlier studies [23,27,29]. A complete restoration of memory functions after shunt insertion was described in a case study with three patients and hydrocephalus by Ogden [25] in only one single patient.

4.2. Secondary deterioration of cognitive functions

The consistent worsening of the alertness score shortly after lumbar puncture as registered in the present patients must be seen with caution, because this result is confounded with a methodological artifact. Despite the substantially slowed simple reaction time all patients with hydrocephalus reacted disproportionately better in the signalled reaction time task. However, after LP the simple reaction time improved substantially and therefore, the numerical difference between simple and signalled reaction time decreased. As a consequence, the alertness score which is defined by this difference worsened. A substantial deterioration in the divided attention task occurred in three patients shortly after lumbar puncture which persisted at follow-up in one. The reason for

Table 4
Neuropsychological performance of patient G.R. (case 4).

Neuropsychological test used	Before lumbar puncture	45 min. after lumbar puncture	Sign. difference	Follow-up	Sign. diff. 1st assessment/follow-up
Finger Tapping test right	13.00 [†]	21.00 [†]	a	17.25 [†]	a
Finger Tapping test left	10.75	27.50	a	17.25	a
d2 concentration speed ¹	96	104	+	114	+
d2 concentration errors ¹	93	107	+	117	+
Benton test ²	7	7	0	8	0
Recurring Figures test ³	43	46	0	41	0
Recurring Words test ³	46	50	0	48	0
IST Memory ¹	102	105	0	116	+
Simple reaction time (ms) ⁴	307	236	+	471 [†]	–
Signalled reaction time (ms) ⁴	264	226	+	309	–
Alertness task ³	60	49	–	80	+
Divided attention omissions ³	47	54	0	47	0
Divided attention reaction time ⁴	741 [†]	656	0	690	0
Go/NoGo task errors ³	20 [*]	46	+	41	+
Go/NoGo task reaction time ⁴	410	380	0	432	0

^a For statistical reasons no significance testing possible.

¹ Standard scores.

² No. of correct reproductions.

³ T-scores.

⁴ Median reaction time in milliseconds (ms). Specification of signs: – significantly worse ($p < 0.05$); + significantly better ($p < 0.05$); 0 no significant difference.

^{*} Marks a cognitive deficit.

Table 5
Time course of life quality and depression.^a

	Patient K.A. (case 1)			Patient T.C. (case 2)			Patient M.F. (case 3)			Patient G.R. (case 4)		
	1st testing	Follow-up	Sign. change	1st testing	Follow-up	Sign. change	1st testing	Follow-up	Sign. change	1st testing	Follow-up	Sign. change
ALQI Social score ¹	16	1	+	5	4	0	14	4	+	20	34	–
ALQI Physical score ¹	19	8	+	3	5	0	16	0	+	21	21	0
ALQI Total score ¹	35	14	+	8	4	0	30	13	+	55	69	–
ALQI Proxy Social score ²	19	1	+	8	4	0	10	0	+	22	26	0
ALQI Proxy Physical score ²	28	5	+	5	8	0	8	0	+	24	16	+
ALQI Proxy Total score ²	47	10	+	13	4	+	18	0	+	58	53	0
Hamilton Depression Rating	24 [*]	13 [*]	+	8	4	0	9	3	+	8	5	0
Beck Depression Inventory	20 [*]	8	+	7	2	0	10	1	+	15 [*]	16 [*]	0

^a Lower scores mean a better quality of life and a lower degree of depression.

¹ Aachen Life Quality Inventory Self-rating version.

² Aachen Life Quality Inventory Proxy-rating version. Specification of signs: – significantly worse ($p < 0.05$); + significantly better ($p < 0.05$); 0 no significant change.

^{*} Substantial depression.

this adverse short-term effect of LP remains as of yet unclear. However, the persistent worsening of two of our patients who both received a shunt in respect to reaction time and divided attention could possibly be explained by a suboptimal function of the shunt and/or the hypothesis of a further decrease of functional brain parenchyma as a consequence of the initial damage. Persisting chronic cognitive impairments even after successful treatment of hydrocephalus by means of shunt insertion or ventriculostomy have been already described in earlier studies [21,22]. The recovery process after treatment of hydrocephalus seems to be complex including persisting cognitive problems and the possibility of secondary functional deterioration [23,27]. On the other hand, a stable improvement could also be achieved without insertion of a shunt, as demonstrated by patient #3 who was treated by repeated LP but did cognitively well at follow-up.

4.3. Quality of life and depression

The self- and proxy-rated quality of life as assessed by the ALQI improved substantially in three patients until the follow-up. Only the self-rated life quality of the patient G.R., who also remained depressive, worsened significantly. On the other hand, according

to the perspective of his partner, the physical aspect of his life quality had improved.

4.4. Suggestions for the future

The present exploratory pilot study provides an indication that cognitive tests of fronto-cortical capacity, motor fine-coordination and simple reaction time seem to be particularly sensitive for the short-term effects of LP. We recommend for the future prospective studies including several follow-up exams e.g. every three or four months for up to two years in order to depict the time course of functional restoration. It would be informative to study also a group of patients whose MH has been treated by repeated LP. In addition, future studies should report also the CSF pressure after the lumbar tap immediately prior to the subsequent neuropsychological testing. Furthermore, we suggest the inclusion of dynamic (PET) and anatomic brain imaging by magnetic resonance tomography (e.g. DWI sequences) in order to detect possible morphological correlates of cognitive changes. Although we used parallel tests in those tasks where learning effects are known, it cannot be excluded that repeated testing gives rise to practice effects. However, if functional worsening occurs, it is more convincing in the

light of possible learning effects which cannot be completely controlled for. In contrast to group studies single-patient studies enable the evaluation of disease impact and treatment outcome for every individual patient and, therefore, shed more light on the individual consequences of treatment [16,17]. It would be promising if both strategies were combined in one study. From a practical point of view the patients should be monitored closely at least by a screening of motor fine-coordination (e.g. Finger Tapping), even if they have not received a shunt.

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