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Recovery in hydrocephalic dementia after shunt operation

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SUMMARY Twenty-three patients with hydrocephalic dementia were studied before and after shunt operation, and improvement was found in 12. Before operation, the improved cases showed more symptoms of confabulation, gait disturbance, urinary incontinence, lack of insight, and constructional apraxia. The improvement was also most marked in these symptoms. The significance of general versus specific symptoms, duration, and aetiology is discussed from a differential diagnostic standpoint, and we conclude that the adequate and early diagnosis of hydrocephalic dementia is essential for good outcome after shunt operation, and that psychiatric and psychometric evaluation enable such a diagnosis to be made.

A syndrome of progressive mental deterioration and neurological disturbances, such as unsteady gait and urinary incontinence, has been described as characteristic of so-called normal pressure hydrocephalus (Adams *et al.*, 1965). Patients with this syndrome showed memory failure, disorientation, and psychomotor retardation. In the further work on delineation of the syndrome, also called low pressure hydrocephalus, adult occult hydrocephalus or, with a more neutral term, hydrocephalic dementia (Granville-Grossman, 1971), symptoms such as apathy, emotional indifference, lack of insight (Braham *et al.*, 1969; Messert and Wannamaker, 1974; Patzold *et al.*, 1974), hostility, and aggressiveness (Crowell *et al.*, 1973) have been emphasised. Theander and Granholm (1967) characterised the amnestic dysfunction in patients with hydrocephalus after subarachnoid haemorrhage as resembling that of Korsakoff's psychosis.

Clinical improvement has been reported after ventriculoatrial shunt operation in about 50% of patients with normal pressure hydrocephalus (Salmon, 1969; Salmon *et al.*, 1971; Messert and Wannamaker, 1974). The possibility of a successful cure emphasises the importance of an adequate recognition of the syndrome. The diagnosis has been based almost exclusively on clinical evaluation including neuroradiology and measurement of cerebrospinal fluid (CSF) physiology. Reports on systematic psychometric evaluation for differential diagnosis and for follow-up after shunt operation are scarce (McHugh and Goodell, 1966; Salmon *et al.*, 1971; Collignon *et al.*, 1975). Granholm and Svendgaard (1972) found that acute onset and a clinical picture dominated by dysmnesia predicted good outcome after shunt operation. The definite diagnosis of normal pressure hydrocephalus is still an important clinical problem even when diagnostic procedures such as RISA cisternography and computerised axial tomography scan are available. Problems of differential diagnosis are frequently encountered, especially against other organic dementias, such as Alzheimer's disease and multi-infarct dementia (Coblentz *et al.*, 1973; Earnest *et al.*, 1974; Katzman, 1976). The present report focuses upon psychiatric and psychometric findings in demented patients in whom clinical, neuroradiological, and CSF findings indicated hydrocephalus. Our aim was to examine whether there are specific psychometric dysfunctions and psychiatric disturbances within the symptom pattern of hydrocephalic dementia. If so, are patients with a preponderance of such characteristics more liable to improve after shunt operation?

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Patients and methods

Twenty-three patients (14 men and nine women) with hydrocephalic dementia, who later underwent

ventriculoatrial shunting, were selected from patients referred to the Psychiatric Clinic for psychiatric and psychometric evaluation. The diagnosis of hydrocephalic dementia was based on clinical investigations, including pneumoencephalography and RISA cisternography, performed at the Departments of Neurology, Neurosurgery, and Neuroradiology, University Hospital of Lund. The shunt operations were carried out at the Department of Neurosurgery. The mean age of the patients was 57.3±10.1 years (range 36–70 years), and the mean duration of the disease, which could be estimated in 21 patients, was 2.9±2.7 years (range 0.25–10 years).

A positive or tentative cause of the hydrocephalus was established before the shunt operation in all but five cases. There were five cases with a history of subarachnoid haemorrhage, seven cases with skull trauma, two cases with aqueduct obstruction, and one case with an Arnold-Chiari malformation. Two cases had previously been operated on for pituitary adenoma and ependymoma respectively, and one case had had a meningoencephalitis. Three cases with head injuries had also abused alcohol. In two cases with idiopathic hydrocephalic dementia, necropsy several years after the shunt operation revealed multiple cerebral infarcts. In one case with a history of skull trauma, the postmortem neuropathological investigation showed changes compatible with Alzheimer's disease.

As reference groups, we used one group of age-matched control subjects (Hagberg and Ingvar, 1976) and three groups of patients with presenile dementia—groups C 2 and C 3, and patients with Alzheimer's disease. Group C 2 with a selective memory disturbance and group C 3 with a general cognitive reduction have been presented fully elsewhere (Gustafson and Hagberg, 1975; Hagberg and Ingvar, 1976). The group with Alzheimer's disease contained 17 consecutive cases with this diagnosis, their mean age was 59.6±4.5 years, and the duration of the disease was 4.7±2.0 years.

Psychiatric rating and psychometric testing were carried out a few days before the ventriculoatrial shunt operation, and seven to 10 days after and three to six months after the operation.

PSYCHIATRIC EVALUATION

This was done by one of us (LG) using a formalised rating scale for psychiatric symptoms (Gustafson and Risberg, 1974; Gustafson, 1975; Gustafson and Hagberg, 1975). The symptom ratings have, in other studies of patients with presenile dementia, been validated against regional cerebral blood flow, and with neuropathological findings in deceased patients (Gustafson *et al.*, 1972; Gustafson and Risberg, 1974; Brun and Gustafson, 1976; Gustafson *et al.*, 1977). The patient's behaviour was also rated by the nursing staff and by members of the family using two formalised rating scales. The items of these latter scales were partly identical with that used by the psychiatrist but also included information on the patient's practical abilities.

PSYCHOMETRIC TESTS

Seven psychometric methods measuring intellectual functions most liable for deterioration in organic dementia were used. *Paired associates* measures immediate recall of verbal material (Cronholm and Molander, 1954). Two parallel versions were used. *Memory for design* assesses the spatial perceptive ability and the immediate memory for simple geometric designs. The score analysis is presented in terms of error points (Graham and Kendall, 1960). *Visual retention test* is considered to test conceptual memory (Benton, 1963). It uses more figures per trial than memory for design, but the figures are simpler. Three parallel versions were used. *Vocabulary* was tested using conventional word definition test (Husen, 1956). *Block design* aims to measure spatial performance as well as inductive capacity. The present version is based on the block design in the Wechsler Bellevue Intelligence Score (Wechsler, 1958). *Colour word test*, a version of the Stroop Color word test was used here to assess cognitive flexibility and intellectual speed. The time for reading the printing-colour which is incongruent with the word is measured (Smith and Nyman, 1959). *Reaction time test*, simple and choice, measures speed of a motor response to visual stimuli (light) either single or as a choice between three lights.

All tests were given before operation. The visual retention, colour word, and reaction time tests were given again immediately after operation, and all tests except memory for design and vocabulary were repeated at follow-up.

The degree of cognitive reduction relative to the estimated premorbid level is rated on a four point scale: (1) mild reduction=preserved verbal ability and dysfunction in a single test not exceeding 2 stanine; (2) moderate reduction=preserved verbal ability and dysfunction in a single test exceeding 2 stanine; (3) marked reduction=dysfunction in all tests exceeding 2 stanine; (4) severe cognitive reduction=verbal reduction with anomia or aphasia. The patients cannot copy memory for design and/or visual retention test patterns.

Results

Considerable clinical homogeneity was found in the 23 cases. The preoperative clinical picture was dominated by loss of vitality, weariness, emotional deterioration, memory failure, confabulation, disorientation, gait disturbance, and urinary incontinence. The patients showed a remarkable lack of insight, sometimes best described as neglect, about their alarming symptoms. The slow, tired, and disinterested appearance of the patients could suddenly change to irritability and dysphoria when they were provoked by questions or other demands. The clinical findings before and after shunt operation are presented in Table 1 together with results from previously studied patients with Alzheimer's disease.

Table 1 *Psychiatric symptoms in hydrocephalic dementia before and after shunt operation, and in Alzheimer's disease. The number of patients with strong/slight evidence of the symptom is expressed as a quotient*

Psychiatric symptoms	*Hydrocephalic dementia (n=23)*		*Alzheimer's disease (n=17)*
	Preoperative	*Postoperative*	
Euphoria	4/3	3/3	2/2
Inertia	14/9	6/14	7/8
Emotional unconcern	10/7	4/8	14/3
Anxiousness, depression	2/3	0/3	6/5
Restlessness	1/6	2/6	3/6
Disorientation	10/6	3/6	9/8
Amnesia for recent events	15/6	6/6	17/0
Amnesia for remote events	4/10	2/8	14/3
Confabulation	8/9	3/5	0/4
Fantastic confabulation	3/0	0/1	0/0
Apraxia	2/5	3/3	9/6
Gait disturbance	12/6	2/11	1/3
Urinary incontinence	8/5	4/4	2/1
Dysarthria	3/3	3/2	4/3
Expressive aphasia	3/6	4/4	13/2

Before operation the patients with hydrocephalic dementia showed inertia, emotional unconcern, disorientation, and amnesia for recent events, which are similar to the symptoms in the cases of Alzheimer's disease. Confabulation, gait disturbance, and urinary incontinence were more frequently observed in the group with hydrocephalic dementia while amnesia for remote events apraxia, expressive aphasia, anxiousness, and restlessness were more common in the Alzheimer's disease group. Clinical improvement was found in 13 cases, while 10 patients appeared unchanged or somewhat deteriorated after the shunt operation. Table 2 shows the patient's preoperative test performance as test results from the four reference groups.

All patient groups deviated significantly from the control group. Moreover, there were significant differences between the performance of the patients with hydrocephalic dementia and those with presenile dementia. The patients with hydrocephalic dementia showed neither the focal memory dysfunction of group C 2, nor the general cognitive dysfunction observed in group C 3. The hydrocephalic patients showed a marked dysfunction of inductive spatial ability with a lower performance in block design. This dysfunction is also obvious in spatioperceptual ability and spatial memory (memory for design plus visual retention

Table 2 *Test performance in hydrocephalic dementia before operation, in patients with Alzheimer's disease, in patients with moderate degree of presenile dementia (C 2 and C 3, Hagberg and Ingvar, 1976), and in an age-matched control group*

Test		*Control group*	*C2*	*C3*	*Hydrocephalic dementia*	*Alzheimer's disease*
	Number:	56	14	16	23	17
	Mean age (yr):	55	55	56	57	60
Vocabulary	Mean	27.4	32.0	26.0	25.7	6.0
	SD	10.9	11.2	13.7	13.9	7.8
Paired associates	Mean	16.5	11.2	11.7	8.8	0.2
	SD	4.6	5.3	5.9	7.1	0.8
Memory for design	Mean	2.8	7.2	10.1	8.5	24.0
	SD	2.6	6.2	6.4	7.5	26.9
Visual retention						
Right	Mean	6.2	5.3	3.0	3.2	−0.2 (n=2)
	SD	1.6	2.2	1.5	1.6	0.9
Wrong	Mean	6.4	7.9	12.0	12.2	18.3 (n=4)
	SD	3.2	4.5	3.7	4.5	4.0
Memory for design and visual retention (number of patients)						
Can copy		0	0	1	6	9
Cannot copy		0	0	0	3	19
Block design	Mean	21.0	19.9	9.5	5.7	0.8
	SD	6.8	5.8	4.9	7.1	2.3

tests) where 22% of the hydrocephalic dementia patients could not reproduce and 13% were unable to copy simple geometrical figures. The patients with Alzheimer's disease of approximately the same age performed extremely poorly on all tests including the verbal one, and did not show the test profiles of the other three patient groups.

Nine (39%) of the 23 patients with hydrocephalic dementia showed an improved cognitive level after shunt operation, seven (30%) patients appeared unchanged, and seven (30%) patients showed a further cognitive reduction postoperatively. A comparison between psychometric and psychiatric evaluation of the postoperative outcome is shown in Table 3. There was good agreement between the two independent evaluations, and disagreement in only three cases. Two of these cases were found to be more alert in the test situation even if their performance did not improve, and were then grouped with the improved patients. This gave a twofold classification with 12 improved and 11 unimproved cases. These two outcome groups were compared with regard to background variables, psychiatric symptoms, and psychometric performance before treatment.

Table 3 *Outcome of operation in hydrocephalic dementia as measured by psychometric tests and psychiatric evaluation*

		Psychometric evaluation	
		Improved	*Unimproved*
Psychiatric evaluation	Improved	10	3
	Unimproved	0	10

No significant difference was found as to mean age at the investigation or as to the preoperative degree of reduced cognition. However, the duration of the disease was significantly shorter in patients who improved (mean=18.5 months) compared to unimproved patients (mean=57.0 months) ($P<0.005$, one-tailed t test; McNemar, 1959).

Table 4 shows the distribution of psychiatric symptoms in the two outcome groups before and after shunt operation. Confabulation, gait disturbance, urinary incontinence, and emotional unconcern were more prominent before operation in patients who improved. On the other hand apraxia, expressive aphasia, and dysarthria were somewhat more frequent in the unimproved cases. Thus, the unimproved cases were more similar to the Alzheimer's disease group than the improved cases. Most patients with hydrocephalic dementia were capable of writing their own names. Only two patients, both belonging to the unimproved group, showed dysgraphia. The manifestation of symptoms before and after operation was compared within the outcome groups. Inertia, emotional unconcern, disorientation, amnesia for recent events, confabulation, and gait disturbance were significantly reduced in the improved cases after operation. The decrease of urinary incontinence was almost significant (Fisher's exact probability test). No significant symptom changes were found in the unimproved group.

Table 4 *Psychiatric symptoms before and after shunt operation of hydrocephalic dementia in improved and unimproved cases. The number of patients with strong/slight evidence of the symptom is expressed as a quotient*

Psychiatric symptom	*Improved cases (n=12)*		*Unimproved cases (n=11)*	
	Before operation	*After operation*	*Before operation*	*After operation*
Euphoria	3/1	2/1	1/2	1/2
Inertia	9/3‡	1/8	5/6	5/6
Emotional unconcern	7/2†	1/3	3/5	3/5
Anxiousness, depression	2/2	0/2	0/1	0/1
Restlessness	0/4	0/4	1/2	2/2
Disorientation	6/3*	0/2	4/3	3/4
Amnesia for recent events	8/3*	1/2	7/3	5/4
Amnesia for remote events	2/6	0/3	2/4	2/5
Confabulation	6/3*	0/2	2/6	3/3
Fantastic confabulations	2/0	0/0	1/0	0/1
Apraxia	0/2	0/1	2/3	3/2
Gait disturbance	8/3‡	0/7	4/3	2/4
Urinary incontinence	5/4	1/3	3/1	3/1
Dysarthria	0/1	0/0	3/2	3/2
Expressive aphasia	0/3	0/2	3/3	4/2

Significance levels (Fisher's exact probability test) indicate difference between preoperative and postoperative ratings as follows: $P<0.025$*, $P<0.01$†, $P<0.005$‡.

Table 5 summarises the psychometric findings before and after operation, and the differences after operation in the two outcome groups. The performance of the control group is given for comparison. Both outcome groups show a stable pattern in the preoperative and postoperative test performance. They show a retained verbal ability, a slight but significant memory reduction, and above all, a marked deterioration of spatial memory and spatioperceptual ability (memory for design and block design tests). The positive change in the improved group was comparatively small and most marked in spatioperceptual ability ($P<0.001$) but could also be found for immediate memory ($P<0.01$).

Discussion

The selection of cases of hydrocephalic dementia for operation was based on neurological evidence of hydrocephalus including pneumoencephalogra-

Table 5 *Preoperative and postoperative test performances in improved and unimproved patients with hydrocephalic dementia*

Test	*Control group (n=54)*	*Hydrocephalic dementia*					
		Improved (n=12)			*Unimproved (n=11)*		
		Preoperative	*Postoperative*	*Difference*	*Preoperative*	*Postoperative*	*Difference*
Vocabulary	27.4/10.9	20.0/15.4			24.9/16.4		
Paired associates	16.4/4.6	6.9/5.5	13.2/6.2	6.3/6.3*	9.2/9.7	7.4/10.6	−1.8/3.5
Visual retention							
Right	6.2/1.6	2.8/1.8	4.0/2.1	2.0/1.9	3.3/2.0	3.4/1.5	−0.4/1.5
Wrong	6.4/3.2	11.1/6.1	9.4/4.2	−2.9/7.3	11.7/4.8	11.6/3.8	0.8/2.7
Memory for design	2.8/2.6	7.3/8.0			6.8/6.7		
Visual retention and memory for design (number of patients)							
Can copy	0	3	1		3	0	
Cannot copy	0	1	0		3	6	
Block design	21.0/6.8	7.1/7.7	12.7/7.4	5.6/3.2†	4.1/6.5	5.1/6.0	1.0/3.9

Differences tested with one-tailed t test, H_0, $m \neq 0$.
*=$P < 0.01$, †=$P < 0.001$.

phy, RISA cisternography, and CSF measurements, and was made regardless of the psychiatric and psychometric findings. In several aspects our series of cases resembles previously published series. The aetiologies were heterogeneous. The material contained idiopathic and obstructive as well as communicating hydrocephalus. There was almost total agreement between the psychiatric and psychometric evaluations of outcome and in agreement with previous reports, about 50% of the patients improved. It should also be pointed out that the results presented are derived from a rather small number of cases.

Before the shunt operation our patients manifested a general mental deterioration in addition to a symptom constellation more specific for the group. They showed cognitive, emotional, and conative-intentional defects such as disorientation, amnesia, emotional unconcern, lack of insight, psychomotor retardation, and inertia. The strong confabulation, the peculiar gait disturbance, and the early appearance of urinary incontinence during the course of the disease seem to be a rather specific symptom pattern in the group. Psychometric tests showed general cognitive reduction including memory dysfunction but with a retained verbal ability. Most specific in this group of patients seemed to be the marked disturbance in perceptual performance and probably also in inductive reasoning. Most likely this is a manifestation of constructional apraxia (Warrington, 1969). Similar findings have been reported in one of the few psychometric studies of hydrocephalic dementia (Collignon *et al.*, 1975).

The emotional disturbances and the amnesic-confabulatory syndrome showed important clinical similarities to frontal lobe lesions (Petrie, 1949; Rylander, 1949; Luria, 1966, 1973), temporal-limbic lesions, Scoville and Milner, 1957; Ule, 1958; Meyer, 1959; Brierley, 1961; Victor *et al.*, 1961), and lesions of the hypothalamic-diencephalic structures (Ule, 1958; Victor *et al.*, 1961; Victor, 1964; Lishman, 1971). Confabulation is not an uncommon feature in organic dementias (Berlyne, 1972), especially in those with fronto-temporal degeneration such as Pick's disease (Brun and Gustafson, 1978). In these patients confabulation is more often accompanied by euphoria, disinhibition, increased tempo of speech, and expressive speech disturbances (Mansvelt, 1954; Escourolle, 1958; Robertson *et al.*, 1958; Gustafson *et al.*, 1978) while other motor functions remain undamaged. The symptom pattern of reduced speed of motor and intellectual performance, urinary incontinence, and a gait disturbance resembling Bruns' frontal lobe ataxia (Bruns, 1892) had also been connected with frontal predilection of ventricular enlargement (Adams *et al.*, 1965; Benson *et al.*, 1970; Mathew *et al.*, 1975).

As seen in Table 4, our patients with hydrocephalic dementia did not have aphasic, agnostic, and apractic disturbances to the same extent as patients with Alzheimer's disease. This may reflect a less severe dysfunction of the temporo-parieto-occipital association cortex in hydrocephalic dementia than in Alzheimer's disease (Critchley, 1953; Luria, 1973; Brun and Gustafson, 1976). In Alzheimer's disease the changes of muscular tension and the small step gait might sometimes cause difficulty in differential diagnosis (Sjögren *et al.*, 1952; Pearcy, 1974; Brun and Gustafson, 1976). However, in Alzheimer's disease the urinary incontinence starts later in the course of the disease.

As has been pointed out in our previous publications on dementia, the clinical picture in hydrocephalic dementia might be clouded by various psychiatric symptoms and reactions such as anxiety, depression, aggressiveness, and dysphoria. This is also the impression from the present study and, in fact, two cases in the best outcome group were first misdiagnosed as conversion reactions with astasia abasia.

Psychometric testing showed a general cognitive reduction with the main dysfunction within spatioperceptual performance (constructional apraxia). The findings are similar to what has been observed in patients with uraemia (Hagberg, 1974) and hepatic cirrhosis (Victor *et al.*, 1965; Rhenström *et al.*, 1974). This might indicate a more diffuse cerebral dysfunction in contrast to the accentuated temporo-parieto-occipital cortical degeneration in Alzheimer's disease (Brun and Gustafson, 1976). The clear difference in cognitive dysfunction between the hydrocephalic group on the one hand and early dementia, C 2 (Hagberg and Ingvar, 1976), and Korsakoff syndrome (Kleinknecht and Goldstein, 1972) on the other also supports this assumption.

Postoperative improvement was found in 50% of the hydrocephalic dementia group. The best therapeutic effect was found in patients with the specific symptom pattern. The improvement was most evident in the specific psychiatric symptoms and psychometrically in a better spatioperceptual performance. It might be justified to assume that the specific symptoms are more directly related to the hydrocephalus and that the less consistent symptoms might be related to other factors such as concomitant injuries to different brain structures (Granholm and Svendgaard, 1972), the aetiology of the hydrocephalus, or the patient's premorbid personality. Therefore positive effect of a shunt operation is more likely when there is a well-defined syndrome of hydrocephalic dementia.

There were no differences between the two outcome groups as to age or degree of cognitive reduction before operation. By contrast, the duration of the disease seems to be of great importance. The improved patients showed more specific symptoms and shorter duration than the unimproved cases. If both outcome groups represent one clinical entity, this finding might indicate a symptom change with time. The emotional reactions and the confabulation observed early in the illness might reflect the adaptive functional capacity of preserved structures (Gustafson and Hagberg, 1975). The clinical differences between the two outcome groups must also be considered against the differences in aetiology. Subarachnoid haemorrhage, obstructive hydrocephalus, and previous intracranial operation were more frequent in the improved group while idiopathic hydrocephalus and skull trauma, often in combination with alcohol addiction, were more frequent in the non-improved cases. This might, to some extent, give the differences in duration of the disease. Presence of a possible cause of hydrocephalus and a complete obstruction might lead to an earlier investigation and detection of hydrocephalic dementia.

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