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Intracranial Arterial Aneurysms in Children and Adolescents

By

O. Heiskanen and **J. Vilkki**

Summary

This is a report on 32 patients with subarachnoid haemorrhage caused by rupture of an intracranial arterial aneurysm in the 0–19 age group. Sixteen of the aneurysms were situated in the bifurcation of the internal carotid artery, 8 in the anterior communicating artery, 4 in the posterior communicating artery, two in PICA, one in the middle cerebral artery, and one in the pericallosal artery.

Direct intracranial clipping of the neck of the aneurysm was carried out in 26 cases, proximal clipping in three, wrapping in two, and common carotid ligation in one case.

There was one death from surgery. The surgical mortality was thus 3.1%. In 80% of cases the result was good, *i.e.*, the patient had no neurological deficit and was able to work or continue at normal school.

The psychological tests indicated some cognitive deficits in aneurysm patients as compared to the control patients. The common feature of these deficits seemed to be an impairment of active retrieval and searching of memory, while common cognitive skills were usually preserved. Four of the five patients who were disabled had obvious deficits in their performances. In the other patients these deficits did not affect their capacity for work or studies, and they were usually unaware of them.

Children seem to tolerate surgery better than adults. Comatose or drowsy patients should not be operated on. The operative method of choice is direct intracranial clipping of the neck of the aneurysm.

Keywords: Cerebral aneurysm; subarachnoid haemorrhage.

Ruptured intracranial arterial aneurysms are rare in the first two decades of life. The percentage of children and adolescents in the large published series varies from 0.5 to 4.6% [7, 10–12, 15]. In 1968–1979 we treated 1,202 patients with arterial aneurysms. Thirty-eight of these patients were in the age group 0–19 years (3.1%). The purpose of the present study is to investigate the results of surgery and the late prognosis in children and adolescents.

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Material and Methods

Patients. Our hospital serves an area with a total population of about 2.1 million and receives all aneurysm patients referred for operative treatment from that area. Our series consists of 32 patients who had undergone surgery, 17 boys and 15 girls. Only two of the patients were under 10 years of age—3 months and 4 years. Ten patients were from 11 to 15 years, and 20 patients from 16 to 19 years of age. The follow-up time varied from 6 months to 11 years. The mean follow-up was 6 years.

Table 1. *Location of Aneurysms in Present Series and in a Series of Adult Patients from this Clinic Studied with Four-Vessel Angiography* [3]

Site of aneurysm	Juveniles		Adults	
	No.	%	No.	%
Carotid bifurcation	16	50	3	2.5
Anterior communicating	8	25	29	24.4
Posterior communicating	4	12.5	28	23.5
PICA	2	6.3	2	1.7
Middle cerebral	1	3.1	40	33.6
Pericallosal	1	3.1	5	4.2
Basilar	—	—	8	6.7
Anterior cerebral	—	—	2	1.7
Posterior cerebral	—	—	2	1.7
Total	32	100	119	100

Site of the aneurysms. Half of the aneurysms were situated at the bifurcation of the internal carotid artery (Table 1). Eighty-eight per cent of the aneurysms were situated in the internal carotid and anterior communicating arteries.

Treatment. Thirty-two patients were operated on (Table 2). Four patients underwent surgery during the first week after the haemorrhage, 11 patients during the second week, 10 during the third week, and 7 later on. There was one operative death. The neck of the aneurysm was clipped intracranially in 26 cases, proximal clipping was done in 3 cases, common carotid ligation was performed in one, and in two cases a wrapping procedure was used. An operating microscope has been in use since 1975, and was used in 10 cases. Two patients developed hydrocephalus after the bleeding and surgery and required a shunt.

In addition we treated six patients conservatively during that time. Two patients were comatose on arrival and died three days and six days after admission without regaining consciousness. Three patients were admitted more than two months after haemorrhage. In line with our policy at that time they were not operated on at so late a stage after bleeding. One patient had not had a subarachnoid haemorrhage, and was investigated for epileptic fits.

Clinical examination. All survivors in the series of 32 surgically treated patients were invited for a personal check-up. One patient refused to attend but answered the questionnaire and stated that he was well, had no subjective complaints, and was fully able to work. Special attention was paid to possible neurological deficits and their effect on the patient's working capacity and the patient's education in comparison with that of the siblings and parents. The patients were asked if the illness and the operation had prevented them from completing schooling or studies or from choosing occupations they desired.

Psychological examination. Twenty-five aneurysm patients underwent psychological examination. Two children under 12 years of age were excluded,

Table 2. *Method of Surgical Treatment and Outcome in Aneurysms of Different Locations*

Site of aneurysm	Method	Outcome			
		Good	Fair	Poor	Dead
Carotid bifurcation	Intracranial clipping	14	1	1	—
Anterior communicating	Intracranial clipping	3	—	1	1
	Proximal clipping	—	—	1	—
	Wrapping	—	—	1	1 *
Posterior communicating	Intracranial clipping	3	—	—	—
	Common carotid ligation	1	—	—	—
PICA	Intracranial clipping	1	—	—	—
	Proximal clipping	1	—	—	—
Pericallosal	Proximal clipping	1	—	—	—
Middle cerebral	Intracranial clipping	—	—	1	—
Total		24	1	5	2

Good = no neurological deficit, normal working capacity, normal school.
Fair = slight neurological deficit, normal working capacity, normal school.
Poor = severe neurological deficit, disabled.
* Died of rebleeding two months after wrapping procedure.

three patients refused, and two were dead. A control group was selected from patients treated at the Clinic of Orthopaedics and Traumatology for arm and leg injuries but had no head injury, recent or old. Fifteen control patients, six women and nine men, were examined with psychological tests. One patient selected for the control group refused. The mean age was 21 years (range 14–27) in the aneurysm group, and 22 years (15–27) in the control group. Thirteen patients in the aneurysm group and ten in the control group had at least high school education. Three aneurysm patients and two control patients were college graduates.

Intellectual capacity was studied with the similarities and block design subtests of the Wechsler adult intelligence scale (Wais)[19] and with sets A, B, and C of Raven's progressive matrices[13]. We expected, however, that intellectual skills would not be as sensitive to the effects of a ruptured aneurysm and its surgical treatment as attention and short-term memory. These aspects of cognitive activity were studied with a modified token test[6, 20], a face matching and recognition test [20], and a visual search for parallels [8]. The immediate memory span was studied with the digit span subtest of the Wais [19], and with rote learning of a 10-word list and with repetition and recall (after homogenous interference) of series of words, sentences, and stories [9].

Benton's visual retention test [4] (form C) was presented in a modified way. The patient was shown each card for 10 seconds and was asked to draw the figure after its removal. After the immediate reproduction of two consecutive designs the patient was asked to redraw the first and then the second of the previous designs. The numbers of correct immediate and delayed reproductions of the 10 designs were counted to form two separate scores.

A test of incidental memory for word pairs was included in the similarities test. The first 10 questions were presented to all patients. They were then asked to recall as many questions as they could, in any order. The number of correct words was the score (max. 20). Then the first word of each pair was given as a cue, and the patient was asked to recall the other word. The number of correct words was the score for cued recall (max. 10).

Incidental memory for colours was studied with set A of the coloured progressive matrices [14]. After the patient had performed the first six matrices, the achromatic (standard) matrices were presented to him. Starting from the sixth back to the first matrix the patient was asked to recall the colours of the matrices. The rest of the matrices (7–12) were presented in the same way. The number of correctly recalled colours was the score (max. 24).

Results

There was one death from surgery (operative mortality 3.1%). One of the patients in whom a wrapping procedure was carried out died from rebleeding two months after the operation. Twenty-four of the survivors had no neurological deficit, were fully able to work, and stated that the haemorrhage and operation had not prevented them from completing their school or studies or restricted their choice of occupation. One patient had a slight residual weakness on the left side which did not, however, affect her working capacity.

In five patients the result was poor. One of them had a psychiatric illness before the bleeding which had prevented him from completing even elementary school. He was disabled by the same disease after the operation. Two patients with anterior communicating, one with middle cerebral, and one with carotid bifurcation aneurysms were disabled because of mental changes, deterioration of intellectual capacity, and epilepsy.

In the psychological examination the aneurysm patients had a lower mean score in the similarities test than the control patients (Table 3). The average time taken to name the 20 figures of the token test was longer in the aneurysm group (47 ± 12 seconds) than in the control group (39 ± 7 seconds). The difference between the groups was significant ($t = 2.34$, $df = 38$, $p < 0.05$).

No marked differences between the groups were seen in the number of correct responses in Raven's progressive matrices (Table 4), in the speed and accuracy of visual search for parallels or faces, in the speed and accuracy to follow the verbal commands of the token test, or in the immediate memory span for sequences of digits or cubes.

In the rote learning of the 10 words, 17 of the aneurysm patients but only 3 of the 15 control patients, failed to recall all the words

Table 3. *Incidental Memory Test Scores*

	Aneurysm N = 25 Mean	S.D.	Control N = 15 Mean	S.D.	t	p<
Word pairs of the *Similarities*						
Free recall	10.1	3.5	12.3	2.5	2.09	0.05
Cued recall	7.4	1.8	8.2	1.2	1.54	N.S.
Recall of colours of *Progressive Matrices*	13.3	3.8	15.7	2.9	2.02	0.05

Table 4. *Intelligence Test Scores*

	Aneurysm N = 25 Mean	S.D.	Control N = 15 Mean	S.D.	t	p<
Similarities (Wais) *	9.9	1.5	11.8	2.8	2.93	0.01
Block Design (Wais) *	9.9	3.1	11.3	3.6	1.35	N.S.
Progressive Matrices **	27.3	4.5 ***	29.2	4.8	1.25	N.S.

* Scaled scores.
** No. of correct responses in sets A, B, and C.
*** One aneurysm patient refused to perform the test; N = 24.

in any of the six consecutive trials. The differences between the groups were significant ($chi^2 = 6.63$, df = 1, $p < 0.01$).

All the patients were able to repeat immediately the two series of three unrelated words in the order they were given. The patients were then asked to recall the first series after repetition of the second, and to recall the second series after recall of the first. Seventeen of the aneurysm patients, but only four of the control patients, needed more than one trial to recall the two series correctly after such homogenous interference. The difference between the groups was significant ($chi^2 = 4.87$, df = 1, $p < 0.05$). The groups did not differ markedly in similar repetition and recall of sentences or stories.

In the visual retention test, the number of correct delayed reproductions was lower in the aneurysm group than in the control group (Table 5), but the accuracy of face recognition was equal in the aneurysm and control groups.

In the incidental memory test the free recall of word pairs of the similarities and recall of colours of the progressive matrices were less accurate in the aneurysm group than in the control group (Table 3).

The patients with left-sided aneurysms did not differ from those with right-hand side aneurysms in any of the test performances. No definite relationship was seen between the site of the aneurysm and the test results.

Table 5. *Visual Retention Test Scores*

	Aneurysm N = 25		Control N = 15			
	Mean	S.D.	Mean	S.D.	t	p <
Immediate reproduction	6.8	2.1	7.7	1.4	1.36	N.S.
Delayed reproduction	5.4	2.4	7.0	1.5	2.29	0.05

Discussion

The sites and sizes of ruptured aneurysms are unusual in children under 10 years of age. This was true of our two patients in this age group. The middle cerebral aneurysm was situated in a distal branch of the middle cerebral artery. There was no evidence of an inflammatory origin of the aneurysm. The other patient had a fusiform aneurysm of the pericallosal artery.

In childhood, subarachnoid haemorrhage due to a ruptured aneurysm seems to occur mainly in the older age groups from 15 to 19 years of age. The *location of the aneurysms* differs from that of adults. Half of the aneurysms were situated at the bifurcation of the internal carotid artery, a rare site in adults. This has also been a common location in other large series in children [1, 12, 15, 17, 21], except in that of Amacher and Drake [2]. The next most common site is the anterior communicating artery. As we receive all aneurysm patients referred for operation from a certain geographical area, the distribution of sites should be representative. The usual adult locations in the posterior communicating and middle cerebral arteries are rare in children. We had neither multiple aneurysms, nor did we observe other congenital malformations, such as coarctation of the aorta or polycystic kidney, reported to be associated with childhood aneurysms [12, 16].

Direct intracranial clipping of the neck of the aneurysm was carried out in 81% of the patients. There was no *surgical mortality* in patients with carotid bifurcation aneurysms. The only operative death occurred in a patient with anterior communicating aneurysm who was still drowsy and disorientated at the time of operation. The autopsy showed a wide infarction in the left hemisphere contralateral to the surgical approach. The clip was in place, and did not occlude any artery. If the operation had been postponed until the patient's level of consciousness had returned to normal the result might have been better.

The majority of patients 80%, had no *neurological deficits* and were fully able to work or to attend normal school. One patient had a slight residual weakness on one side, which did not affect her working capacity.

The *psychological test results* indicated some cognitive deficits in the aneurysm patients. They performed less well than the control patients in the similarities subtest of the Wais, in the naming task, and in some tests of learning and memory. No marked differences were seen between the groups in the immediate memory span in visual perception recognition and reasoning, or in constructional ability. The cognitive deficits were not statistically related to the side of the craniotomy or to the site of the aneurysm. The common feature of the deficits seems to be an impairment of active retrieval and searching of memory, while recognition and cued recall as well as common cognitive skills in simpler activities were usually preserved. The control group had on an average slightly better education than the aneurysm group, but such a small difference could hardly be the reason for the differences in the test results between the groups.

Four of the five patients unable to work or attend normal school had obvious cognitive deficits. Two of them had deficient performance in the memory tests and in the constructional tasks. One worked slowly and inaccurately. He was drowsy, presumably because of heavy antiepileptic medication. An eleven years old girl (excluded from the statistical analysis) had poor short term memory and showed retardation in reasoning and constructional skills. She was not able to attend normal school. The fifth patient had normal cognitive performance, but he suffered from neurotic tension and fears that were not due to the rupture of the aneurysm. Three of the patients able to work or to attend normal school did the least well of all patients in the memory tests. Thus factors other than cognitive efficiency may be essential determinants of the working capacity.

The *result of surgery* was good in 60% of the adult patients in earlier series from this clinic [18]. It seems that children tolerate the operation better than adults. A poor result seems to occur more frequently in anterior communicating aneurysms, but the number of patients is too small to allow conclusions about the surgical risks in aneurysms at different sites.

Two thirds of the patients with fatal outcome after surgery in Patel's [12] and Sedzimir's [15] series were comatose or drowsy at the time of surgery, as was our single patient with fatal outcome. Thus it appears surgery should be undertaken only if the patient is fully alert and orientated, a recommendation generally accepted for adults. The surgical method of choice is direct intracranial clipping of the neck of the aneurysm. In selected cases, proximal clipping is advisable.

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Authors' address: O. Heiskanen, M.D., Neurosurgical Clinic, Topeliuksenkatu 5, SF-00260 Helsinki 26, Finland.