

Arachnoid cysts in children: a European co-operative study*

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Abstract. The data on arachnoid cysts in children (0–15 years) operated upon between 1980 and 1988 were analysed in a retrospective, co-operative study. The results from 285 patients indicate a predominance of these lesions in boys (64%) more than girls (36%) and a mean age of 6 years at onset of symptoms. Focal EEG patterns corresponding to the cyst's location were encountered in 32%. About 40% of all cysts were located along the midline, the sylvian fissure representing the predominant location. Open surgery, i.e. total excision or marsupialization (together 43.3%), emerged as the first-choice surgical procedure. The type of surgery switched somewhat to shunting procedures in cases of lesions in deeper locations (22.8%). Morphological results on follow-up revealed a reduction of the size of the cyst in a significant majority (61%); in 18% the cyst had disappeared completely on CT scans. There was an obvious correlation between postoperative morphological findings and clinical outcome.

Key words: Arachnoid cysts – Co-operative study – Paediatric age – Clinical-morphological correlation

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Some 150 years after it was made, Bright's original description of arachnoid cysts in 1831 proves to have shown remarkably accurate observation. Bright defined this entity as "serous cysts forming in connection with the arachnoid, and apparently lying between its layers" [9]. In fact, arachnoid cysts are well-defined, easily detectable, benign and in the majority of cases easily accessible lesions, and therefore should present practically no problems to modern, high-standard neurosurgical centres. Bright's first observation has been supplemented by a virtual mountain of communications which reflect the ongoing discussion of aspects of the pathogenesis, diagnosis and treatment. Reports of unique or unusual cases add to our knowledge of the variability of this entity [34, 60, 67, 75]. Occasionally arachnoid cysts were confused with subdural hematomas [53, 82] or other clinical entities [7] in very early reports. In 1913 Zesas and Bachelier presented the first collection of all singular observations by some 38 different authors at that time; in 1923 Demel [15] again reviewed the literature thoroughly and concluded that for the vast majority of patients, favourable results can be expected from surgery such as trepanation, drainage or excision. Frazier [19], calling these lesions pseudotumours, advocated surgery, a strategy followed by increasing numbers of authorities in the decades that followed [1, 27, 51, 62, 71, 72, 76, 86].

For introduction we may simply put down some facts that are agreed upon unanimously. The former terminological confusion ("meningitis serosa circumscripta", "cerebral pseudotumour", "chronic arachnoiditis", etc.) is now at an end: true arachnoid cysts are benign malformations of developmental origin. Other benign cysts to be distinguished are [25]: (1) leptomeningeal cysts caused by sequestration of subarachnoid space by infection, trauma or haemorrhage, comprising parts of the subarachnoid space; (2) porencephalic cysts, which are cavities due to brain parenchyma defects with frequent communication to CSF pathways; (3) neuroepithelial cysts, which appear similar to arachnoid cysts but are lined by ependymal or choroid epithelial cells [20]. For completeness, cystic tumours, chronic subdural haematomas, col-

loid cysts of the III ventricle and hydatid cysts may be mentioned in connection with differential diagnosis.

The ultrastructure of arachnoid cysts has been thoroughly investigated and is distinct from that of normal arachnoid membranes [55, 65]: (1) the arachnoid membrane is split at the margin of the cyst; (2) there are no traversing trabecular processes within the cyst; (3) there is a very thick layer of collagen in the cyst wall, which presumably participates in the collagen synthesis.

The cysts normally contain clear, CSF-like fluid, although in some instances blood breakdown products may be encountered. Such blood fragments usually derive from traumatized blood vessels, which, lying in the cyst wall or even traversing the cyst, are more susceptible to trauma because of their isolated topography [2, 10, 13, 22, 41, 42, 44, 48, 57, 77, 80, 85]. They represent the normal cortical vasculature, with the superficial cerebral veins generally on the outer, convex cyst surface and the arterial branches near the inner surface right on the cortex [3]. Logically, they should be dealt with with the utmost care [29].

Arachnoid cysts are believed to account for about 1% of all intracranial expansive lesions [57]. However, a considerable number may remain undetected, judging from the unsuspected incidence of 5 per 5000 found in a systematic necropsy study [66]; the new imaging techniques are apparently diminishing the gap between actual diagnosis and virtual incidence by providing a significantly higher rate of incidental findings. According to results in larger mixed series (i.e. including both children and adults), 60–90% of patients belong to the paediatric age group, and most arachnoid cysts even become symptomatic in early childhood [4, 21, 30, 31, 33].

Demel and Frazier led the way in a school of thought that considered arachnoid cysts to be a matter for surgery, and still today a convincing majority pleads for craniotomy and excision [3, 11, 12, 17, 23, 25–27, 29, 32, 35, 45, 47, 49, 50, 58, 68, 78, 79]. Mainly in the last decade, however, a significant number of communications have argued for the more simple shunting procedures [14, 18, 24, 28, 30, 31, 36, 38, 43, 46, 61, 63, 69, 70, 73, 74, 84]. The two surgical methods to choose between, the uncertainty when to operate, and the lack of comparable results from numerous communications made us (R.W.O. and J.H.) initiate this cooperative study. It was outlined to include children aged 0–15 years with arachnoid cysts operated upon between 1980–1988. Questionnaire forms were sent to selected institutions in Europe, and 17 institutions from 9 different European countries agreed to join the study (see list).

Clinical material

Two hundred and eighty-five cases of arachnoid cysts in children (0–15 years) have been collected. Interestingly, there is a distinct predominance of boys (182 patients, 63.8%), whereas girls (103 patients) constitute only 36.2% of the patients. A recent head trauma leading to admission had occurred in 47 cases (16.5%). The age distribution (Fig. 1) reveals a significant peak in the first 2 years of life, apparently reflecting a quite common coincidence of the cysts

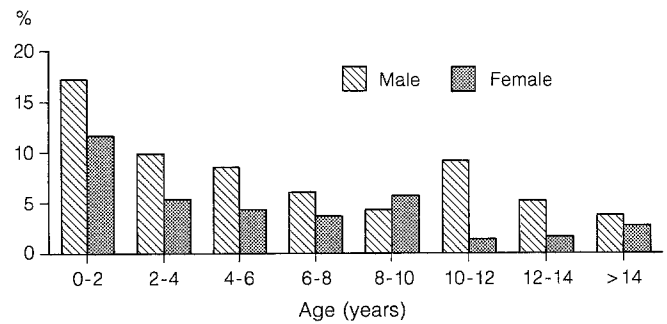


Fig. 1. Distribution of age at onset of symptoms in 285 children with arachnoid cysts

Table 1. Symptoms on admission in 285 children with arachnoid cysts

Raised intracranial pressure	48.8%
Lateralizing symptoms	36.5%
Increased head circumference	30.5%
Mental retardation	20.7%
Seizures	18.2%
Ocular symptoms	14.0%
Cerebellar symptoms	11.9%
Endocrinological abnormalities	4.9%
Speech disorders	1.8%

Table 2. Distribution of locations of arachnoid cysts

Sylvian fissure	34.6%
Cerebello-medullary cistern	15.5%
Cerebral convexity	12.9%
Sellar area	11.3%
Other	10.4%
Interhemispheric fissure	6.5%
Supracollicular area	5.2%
Cerebello-pontine angle	2.3%
Clival and interpeduncular area	1.3%

with symptomatic hydrocephalus. The mean age at onset of symptoms is 6 years.

Signs of elevated intracranial pressure, frequently associated with macrocrania, occur in more than 50% of cases and are the leading symptom on admission (Table 1). The other symptoms are mostly related to the cyst location; seizure disorders are reported in 18.2% of patients. Normal EEGs are found in 19.2%, focal signs corresponding to the cyst are encountered in 31.9%

Morphological appearance

Computed tomography (CT) reveals a circular shape in 37% of cases, a quadrangular in 13%, and an oval shape in 43%; ruptured cysts are found in 19 cases (7%). Altogether, an estimated size over 50 cm³ is found in 44.5%. Such larger cysts are mainly located in the sylvian fissure (44.4%), the cerebello-medullary cistern (13.7%), the sellar region (11.2%) and the cerebral convexity (11.3%). Half of the cases (49.1%) present with displacement of CNS structures; the distribution of different locations is listed in Table 2. By far the most common location is the sylvian fissure (34.6%), followed by the cerebello-medullary cistern (13.7%). Approximately 40% are located somewhere along the midline.

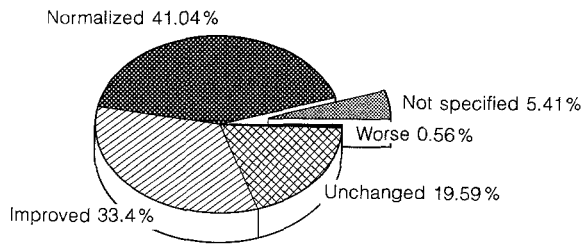


Fig. 2. Status of preoperative symptoms at postoperative follow-up

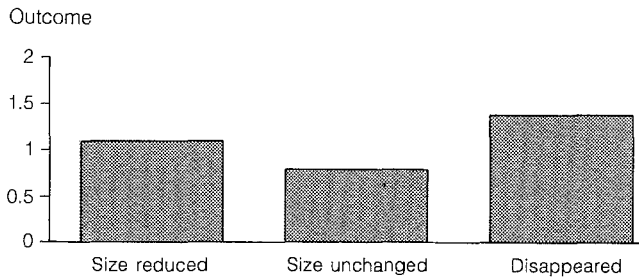


Fig. 3. Correlation of postoperative morphology and clinical outcome

Table 3. Type of initial surgical procedure

Cyst shunting	19.4%
Ventricular shunting	15.1%
Marsupialization	17.3%
Microsurgical total excision	26.0%
Marsupialization and shunt	7.3%
Other procedures	6.7%
None (conservative treatment)	7.9%

Table 4. Correlation between location of cyst and type of treatment

	Deep	Superficial
Ventricular shunting	12.0%	24.0%
Cyst shunting	22.8%	22.0%
Marsupialization	18.5%	26.2%
Microsurgical total excision	33.7%	16.0%
Marsupialization and shunt	8.2%	10.0%
Other procedures	4.8%	1.8%

Table 5. Sellar region cysts: outcome scores after different surgical procedures in 18 patients

Cyst shunting	1.39
Ventricular shunting	0.96
Marsupialization	0.92
Microsurgical total excision	1.18
Marsupialization and shunt	0.58

Treatment

The analysis of the different surgical procedures initially employed (Table 3) reveals an obvious predominance of open surgery (marsupialization and total excision together: 43.3%) over cyst shunting (19.4%). If superficial and deep cyst locations are related separately to the type of surgery, there is a slight shift towards cyst shunting in deep locations (Table 4).

Table 6. Incidence of secondary procedures after excision/marsupialization and cyst shunting

Type of surgery	Initial procedure	Patients with additional procedures		Total no. of additional procedures
	(n)	n	(%)	
Cyst shunting	63	19	30	47
Ventricular shunting	50	28	56	31
Marsupialization	57	11	19	14
Microsurgical total excision	87	6	6.8	6
Marsupialization and shunt	24	6	25	7
Other procedures	22	8	36	8

Follow-up

Except for 24 individuals who were lost to follow-up, all children (261, i.e. 91.58% – the vast majority) had clinical and morphological follow-up for a mean of 38.7 months. In summary, the preoperative symptoms normalized or improved in the great majority (Fig. 2); only 0.56% deteriorated after surgery. In spite of the relatively large number of cases in this series, the evaluation of individual follow-up symptoms did not result in useful statistical data. At CT follow-up, the cyst was no longer visible in only 18%; in 61% it was decreased in size and in 21% it remained unchanged.

With regard to the quantity and variability of symptoms, outcome had to be defined arbitrarily. It is expressed in “mean change in symptoms” for each individual patient, and the following points were assigned to four different qualities: worse 1.5, unchanged 0, improved 1, normalized 2. The mean of the assigned numbers expresses the “mean change in symptoms”, e.g. if one of two symptoms is unchanged and the other improved, outcome is calculated as $(0 + 1)/2 = 0.5$.

Figure 3 demonstrates an obvious correlation between postoperative morphology and clinical outcome. Due to the high variability of symptoms, locations and surgical modalities, however, the overall correlation of these three features did not yield statistically significant data. However, in single locations, as with the example of cysts in the sellar region (Table 5), it might be concluded that shunting procedures appear to be the preferred surgical approach at deep locations [54].

Due to unsatisfactory clinical and/or morphological results, a total of 113 additional surgical procedures were performed in 78 patients, i.e. 27.4%. The need for a second surgical procedure turned out to be more frequent after cyst-shunting (30%) than in the two combined groups of patients who had undergone marsupialization and total excision (25.8%). This difference is significantly aggravated in a comparison of the total amounts of further procedures required: 19 individuals with initial cyst shunting required 47 additional surgical procedures, while the marsupialisation and total excision groups together had 20 further operations in 17 patients (Table 6). Although outcomes in the two groups did not differ significantly as in other series [56], it appears noteworthy at this point that the number of reoperations is more than twice as high in the group with initially shunted cysts.

Discussion

True arachnoid cysts are of developmental origin, without any doubt. Discussion of the two hypotheses – development derangement of the arachnoid accompanying minor aberrations of CSF flow [72, 81] versus agenesis of brain structures resulting in dilatation of CSF-containing spaces to make up for the loss of cerebral volume [6, 57,

64] – is decreasing. The two entities arachnoid cysts and cerebral agenesis may coincide, but such coincidences are considered virtually as rare entities. The agenesis hypothesis is being gradually abandoned, as there is not too much left arguing for this theory – on the contrary: (1) it is impossible to imagine an aplastic temporal lobe reexpanding after surgery; (2) CT calculation of brain volumes does not reveal significant differences in volume between the two hemispheres [47, 78]; (3) there are no corresponding neurological disorders; (4) there is no indication of aplasia of brain structures in arachnoid cysts located other than temporally (e.g. in the midline). Although the theory of aplasia goes on being raised again and again, there seems no reason to continue doing so.

The second question deals with the potential communication to CSF pathways and the mechanism of expansion of arachnoid cysts, which, despite meticulous investigations, remain unclear. Essentially, there are four possible ways for cysts to enlarge: (1) secretion from the cyst wall, which has been indicated by ultrastructural and histochemical findings [25, 55]; (2) a valvular mechanism making for easier ingress into than egress from the cyst [40, 76]; (3) a persisting communication between cyst and CSF pathways, thus maintaining expansion by pressure waves of venous origin during coughing, straining, etc. [83]; and (4) osmosis or filtration processes similar to CSF passage from the subarachnoid space to dural venous sinuses in pachionian granulations [55].

By whatever mechanism, enlargement is usually accompanied by development of clinical symptoms. Arachnoid cysts are thought to be communicating in their earlier stages [55]; at later stages they rather seem to become sealed off. An inverse relationship has been suspected between size of cyst and degree of communication. Different protein concentrations in cyst and lumbar CSF, episodic appearance of clinical symptoms and relative stasis of dyes or traces in the cavity may be considered as evidence of only intermittent communication [23].

The observation of relative stasis again raises the question of the validity of additional neuroradiological investigations in order to establish appropriate treatment decisions. Such investigations appear literally imperative when morphological patterns do not provide clear differentiations, e.g. between enlarged great cisterns and true arachnoid cysts in the posterior fossa [16, 37, 39, 59]. Despite the new imaging possibilities, employment of dynamic studies such as encephalography with positive contrast or isotopes has been suggested [5, 69]. However, the experience of false findings with either only temporary communication or pure filtration motivated a number of authorities simply to give up such rather intricate investigations in children [30, 35]. The potential visualization of flow phenomena on MRI may be considered a sufficient substitute, but this should be used only with reservations as a basis to decide for or against surgery.

In addition to CSF diversion procedures for significant occlusive hydrocephalus due to arachnoid cysts, two operative modalities are mainly employed. According to this study, craniotomy with excision or marsupialization is by far the favourite surgical option. Although usually a fairly easy and appealing procedure from the microsurgical

point of view, it results in some significant operative complications, morbidity [3, 12], and even a low rate of mortality [12, 23, 58, 83]. On the other hand, the apparently safer procedure of cyst shunting is more often followed by secondary surgical efforts and danger of life-long shunt dependency. Although the value of results from retrospective studies is limited, it may be suggested that, with regard to the benign nature of this entity and the potential risk of morbidity and even mortality, deeply located arachnoid cysts should be considered for shunting procedures rather than open surgery. It is widely agreed that the onset of obvious, related symptoms should prompt a decision for surgical treatment. Bret and Benes [8] advocate surgery right after the first appearance of symptoms and correlate this stage with the structural transformation from elasticity to plasticity of brain tissue. Although this appears logical and is shown by the results of this study to a certain extent, it remains questionable whether morphological and functional patterns do necessarily correlate.

The principal surgical goal is to eliminate the expansive and/or obstructive effect of arachnoid cysts. Pure fenestration of only the outer membrane of the cyst in superficial locations appears the easiest and safest procedure but carries a significant risk of reformation of the resected membrane. If CSF pulse waves are allowed to pass the former cavity through sufficiently large windows in a bipolar fashion, this risk appears definitely lower. However, this requires dissection in deeper areas with a potential increase in morbidity. Shunting procedures, in turn, are obviously safer but accompanied by a higher incidence of additional surgical procedures and the threat of life-long shunt dependency. Since neither of these traditional modalities can be considered satisfactory, more recent reports on endoscopic fenestration [52] and/or internal drainage [8, 29] appear to deserve our increased attention.

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Geographic editor's comment

I must compliment the authors on having taken the initiative to start this co-operative study. Most paediatric neurosurgeons do not have the opportunity to see more than a few cases of arachnoid cyst a year; it will take quite a long time before they have extensive personal experience. Such co-operative studies are an excellent way to obtain broader information.

The conclusions in the study are to be recommended. More such co-operative studies should be done on different topics.

Karl H. Hovind, Vetre