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Intracranial arachnoid cysts: What is the appropriate surgical technique? A retrospective comparative study with 61 pediatric patients

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ABSTRACT

Objective: Symptomatic intracranial arachnoid cysts (ACs) should be treated either through microsurgical (MS) or endoscopic surgical (ES) fenestration. Implantation of cysto-peritoneal shunt (CPS) system is another treatment option with decreasing indication. In our study, we compared the complication and revision rates between the three operative techniques in pediatric patients.

Methods: We included patients below 18 years with symptomatic intracranial ACs operated between 2004 and 2021. Initial symptoms, location, complication rate, clinical and radiological improvement, postoperative events and revision rate were compared retrospectively.

Results: Sixty-one patients; 33 (54.1%) MS operated (mean age 7.6 years), 18 (29.5%) ES operated (mean age 6.2 years) and 10 (16.4%) with CPS (mean age 3.0 years) were collected. The most common initial symptom was headache in 45.9%. 20 (32.8%) postoperative events were documented. The highest revision rate (60%) was seen in the CPS group compared to 33.3% in MS group and 16.7% in ES group. 31 patients harbored perisylvian ACs, 89% remained event-free after ES, 71% after MS and 20% after CPS. Clinical improvement immediately after surgery was observed in 58 patients (96.9% in MS, 88.9% in ES and 100% in CPS). A radiological volume reduction could be proven postoperative in 51 patients (78.8% MS, 88.9% ES and 90% CPS).

Conclusion: Endoscopic fenestration of AC is a safe and efficient technique which is being widely used nowadays with the highest event free survival compared to microsurgical fenestration especially in perisylvian arachnoid cysts. CPS shows on long terms the highest revision rate but carries the least surgical risks.

1. Introduction

After reviewing a total of 48,417 adult patients who underwent brain magnetic resonance imaging (MRI) over the study period, intracranial arachnoid cysts (AC) were identified in 661 patients (1.4%).[1] In comparison to pediatric population, intracranial arachnoid cysts were present in around 2.6% of 11,738 patients who were 18 years of age with a slight male predominance with ratio 1.8:1[2]. Arachnoid cysts can occur at various locations, most frequently in the middle cranial fossa (49%), followed by posterior fossa (38%). During observation of 111 ACs over 3.5 years, 9.9% showed increase in size, 11.7% diminished, and 78.4% were unchanged.[2] Younger patients at diagnosis are more likely to need surgery,[2] and possibly benefit more in terms of neurological function.[3–5]

In most cases, deciding whether or not to perform surgery for an accidently discovered arachnoid cyst is difficult. However, there is a consensus among neurosurgeons that symptomatic arachnoid cysts should be treated. There are various surgical techniques available, including open microsurgery,[5–11] endoscopic procedures[8,12–15] and shunting procedures.[8,16,17] The key to successful treatment is relieving pressure inside the cyst, which can be achieved through fenestration to subarachnoid spaces or ventricular system or insertion of a cystoperitoneal shunt (CPS). While many recent studies have compared microsurgical versus endoscopic techniques[9,12,18–20] and have found that endoscopy is increasingly favored due to its minimal invasiveness,[21] others have found no significant difference in outcomes or complications between shunting, craniotomy, and endoscopy.[8,19] However, in cases of intraventricular arachnoid cysts, operating via endoscope may provide a clear advantage.[22] Nevertheless, shunting may have more long-term

Abbreviations: AC, Arachnoid cyst; CISS, Constructive interference in steady state; CPS, Cystoperitoneal shunt; CSF, Cerebrospinal fluid; ES, Endoscopic surgery; ICP, Intracranial pressure; MRI, Magnetic resonance imaging; MS, Microsurgery.

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complications which should not be overlooked or underestimated.

The aim of this study is to compare the clinical and radiological courses after surgery to identify if there is a technique superior than the other in pediatric patients.

2. Patients and methods

The study was approved by our ethic committee (S-084/2022). According to this approval, consent of the guardians was not necessary because of the retrospective nature of the study. We reviewed our database retrospectively and performed a comparative mono-centric study including all patients below age of 18 years with symptomatic intracranial arachnoid cysts whom their first surgical treatment took place in our hospital between 2004 and 2021 with a minimum clinical follow-up of 12 months for clinical and radiological data. We analyzed and compared the initial symptoms, cyst location, complication rate, clinical and radiological course, revision rate and hospital stay between the three operative techniques. As the study spanned a 17-year period and involved three different pediatric neurosurgeons, we were able to observe the use of three surgical techniques over time. Through our observations, we noticed that one technique was increasingly favored over the others, likely due to the personal preferences of the surgeons involved.

The cysts were categorized into six locations based on their position: perisylvian, suprasellar, interhemispheric, retrocerebellar, cerebellopontine angle, and other locations. Depending on the treatment modality, patients initially received one of three approaches: microsurgery fenestration (MS), endoscopic surgery (ES), or cystoperitoneal shunt insertion (CPS).

2.1. Surgical techniques

In cases of microsurgical fenestration, a small craniotomy was done overlying the arachnoid cysts, usually without neuronavigation, many fenestrations were achieved to communicate the AC with the nearby subarachnoid spaces or ventricular system. In cases of endoscopic fenestration, a burr hole was planned, using neuronavigation, to allow with one trajectory the fenestration of the cyst to nearby subarachnoid spaces or ventricle itself (Fig. 1). In cases of insertion of cystoperitoneal shunt, a small burr hole was done overlying the arachnoid cyst and a ventricular catheter is inserted after opening the dura and connected to a programmable pressure gradient valve usually without gravitational unit and finally connected to a peritoneal catheter inserted via open technique through a small paraumbilical incision.

2.2. Clinical and radiological outcome criteria

All patients were evaluated for success both clinically and radiologically. Clinical success was defined as an improvement in presenting symptoms following surgery. Radiological success was determined by a decrease in cyst size across all diameters on MR imaging following surgery. To assess the effectiveness and superiority of surgical treatments,

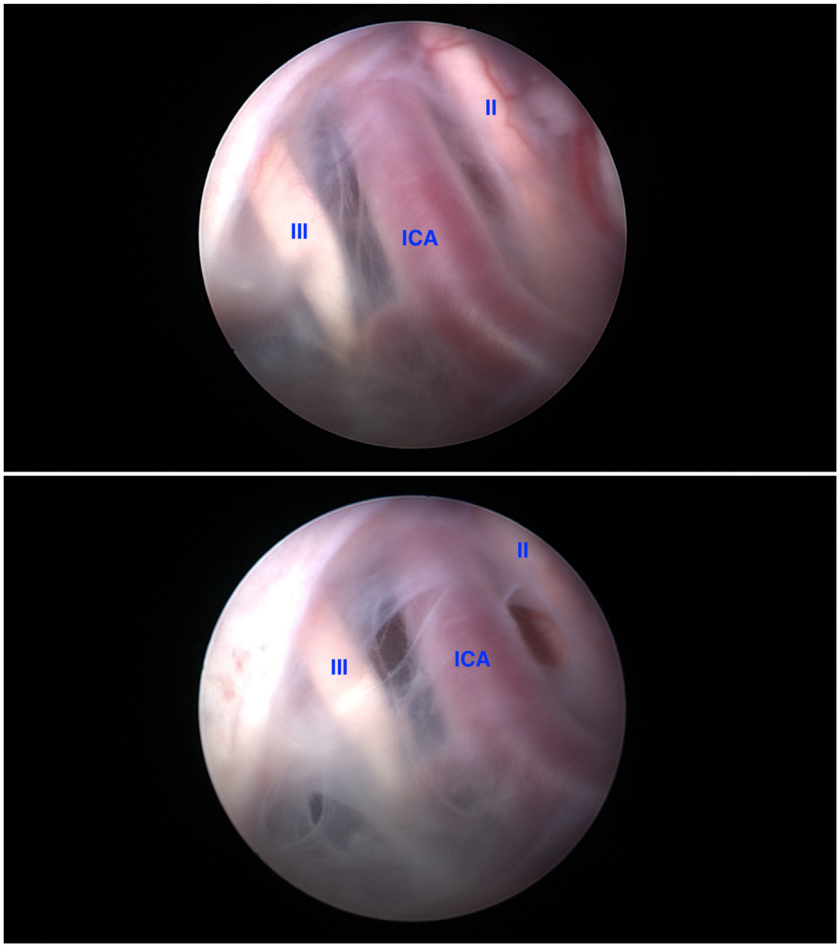

Fig. 1. a: An endoscopic view of the medial wall of a perisylvian arachnoid cyst, **b:** 3 endoscopic fenestrations were done between internal carotid artery, optic nerve and oculomotor nerve as well as lateral to it.

we also analyzed the incidence of postoperative adverse events requiring further surgery at fixed intervals of 6 months, 12 months, and 48 months post-surgery. These events included subdural hygroma or hematoma requiring evacuation, insertion of CP shunt, revision of previously inserted CP shunt, duraplasty, or revision due to wound or implant infection.

2.3. Statistical analysis

Continuous variables were reported as mean and standard deviation. Independent *t*-tests were used for intergroup comparisons of continuous variables. Revision rates between the study groups were compared using Chi square test and Fischer Exact test. The Kaplan–Meier estimator was used to estimate the time to event from lifetime data in cases of need for revision surgery due to adverse events. Two-sided level of significance was set at 0.05. Statistical analyses were performed using SPSS (v27.0, IBM-Corp, Armonk, NY, USA).

3. Results

3.1. Patients' characteristics

A total of 61 pediatric patients were operated for symptomatic intracranial AC for the first time in our hospital in the last 17 years. Thirty-three (54.1%) were fenestrated with MS technique (mean age 7.6 ± 5.98 years), 18 (29.5%) with ES technique (mean age 6.2 years ±5.36 years) and 10 (16.4%) received initially CPS (mean age 2.95 ± 3.96 years), one of them was a cystoventricular shunt in form of a unidirectional valve-guided extracranial stent by a young infant 2 months old with a retrocerebellar AC. The MS group had the longest follow-up period with a mean follow-up of 78.5 ± 62.3 months, followed by the CPS group with a mean follow-up of 77 ± 68.5 months and the ES group with a mean follow-up of 43 ± 41.2 months, see Table 1.

Almost half ($n = 31$; 50.8%) of the ACs were perisylvian, 12 (19.7%) ACs had an interhemispheric location, and the rest showed retro-cerebellar (8.2%), cerebellopontine angle (8.2%), suprasellar (6.6%), location or others, for details see Table 2.

3.2. Preoperative findings and indication for surgery

All patients presented with at least one clinical symptom rather than headache, which primarily indicated the surgery. The most common initial symptom was headache in 45.9%, 27 (44.3%) patients showed at least one or more focal neurological deficits; for example, 8 patients suffered from oculomotor dysfunction, 5 from hemiparesis, 4 showed deterioration of visual acuity, 3 manifested with ataxia. Twenty (32.8%) children showed nausea and vomiting, progressive macrocephaly was found in 31.1% which were mostly younger in age where headache was not commonly clinically evident. In 15 patients (24.6%), there was radiological evidence of size increase during follow-up, 13 children (21.3%) suffered from deterioration of the general condition, for example failure to thrive, in 9 children (14.8%) papilledema was detected as a sign of elevated ICP. Fig. 2 summarizes the initial findings in our cohort.

Table 1
Patients' characteristics, adverse events and events at 6 m, 12 m and 48 m.

n (%)	MS group	ES group	CPS group
No. Of patients	33 (54.1%)	18 (29.5%)	10 (16.4%)
Age (SD[a])	7.6 years (±5.98)	6.2 years (±5.36)	2.95 years (±3.96)
Follow-up (SD)	80.9 months (±62.5)	38.4 months (±38.6)	74.0 months (±71.1)
Postoperative adverse events (%)	1 (3%)	2 (11%)	0 (0%)
Postoperative hygroma (%)	6 (18.2%)	1 (5.6%)	0 (0%)
Events at 6 months (%)	9 (27.8%)	2 (11.1%)	0 (0%)
Events at 12 months (%)	11 (33%)	2 (11.1%)	1 (10%)
Events at 48 months (%)	11 (33%)	3 (16.7%)	6 (60%)

[a] Standard deviation.

3.3. Postoperative clinical and radiological course

Immediate postoperative clinical improvement was observed in 32/33 patients (96.9%) in the MS group, 16/18 patients (88.9%) in the ES group and in 10/10 patients (100%) in the CPS group. A radiological volume reduction of the arachnoid cyst could be proven in a post-operative MRI in 51/61 patients (78.8% MS, 88.9% ES and 90% CPS).

3.4. Event free survival

At the 6-month checkpoint, 9/33 patients (27.2%) in the MS group required additional surgery, compared to 2/18 patients (11.1%) in the ES group and 0% of patients in the CPS group. By the 12-month checkpoint, one more patient in the MS group required surgery, bringing the total to 10/33 patients (30.3%), while the percentage remained the same in the ES group and increased to 1/10 patients (10%) in the CPS group. Four years later, at the final checkpoint, 11/33 patients (33%) in the MS group required revision surgery, compared to 3/18 patients (16.7%) in the ES group. In the CPS group, 6/10 patients (60%) required additional surgery compared to previous checkpoints. Postoperative subdural hygroma was encountered in 7 patients (11.5%), 6 times after MS and one time after ES. One patient suffered from subdural hematoma that required evacuation, mainly after head trauma (Fig. 3). One patient required wound revision due to CSF leak. Partial oculomotor paresis and partial facial paresis were encountered in 2 patients. Totally, we noticed 3 (5%) postoperative surgery-related complications. 2/18 patients (11%) from the ES group needed later a CPS, in comparison to 8/33 (24%) patients from the MS group, $\mathbf{p = 0.46}$.

In total, we encountered 20 postoperative events during follow-up that necessitated revision surgery. One third (11/33) after MS, one sixth (3/18) after ES and 60% (6/10) after CPS. Based on the groups of the various surgical techniques, the CPS group had the highest post-operative revision rate (60%), compared to the MS with 33.3% and ES groups with (16.7%), $\mathbf{p = 0.076}$. See Fig. 4.

3.5. Patients <2 years of age

We encountered 21/61 patients <2 years of age. There was a significant tendency towards CPS in that age group, 7/10 CPS were inserted in children below 2 years of age versus 5/18 patients who received ES and 9/33 patients who received MS were <2 years of age., $\mathbf{p = 0.04}$. Accordingly, event free survival was significantly lower in children below 2 years of age, where only 10/21 patients <2 years remained event

Table 2
AC's locations and related adverse events.

(%)	Peri-sylvian	Inter-hemispheric	Sellar-suprasellar	Location	Cerebello-pontine Angle	Other	Total
				Retro-cerebellar			
No. Of patients	31 (50.8%)	12 (19.7%)	4 (6.6%)	5 (8.2.%)	5 (8.2%)	4 (6.6%)	**61 (100%)**
Postop. Adverse events	1 (3.2%)	0 (0%)	1 (25%)	0 (0%)	1 (20%)	0 (0%)	**3 (4.9%)**
Postop. Hygroma	5 (16.1%)	1 (8.3%)	0 (0%)	1 (20%)	0 (0%)	0 (0%)	**7 (11.5%)**

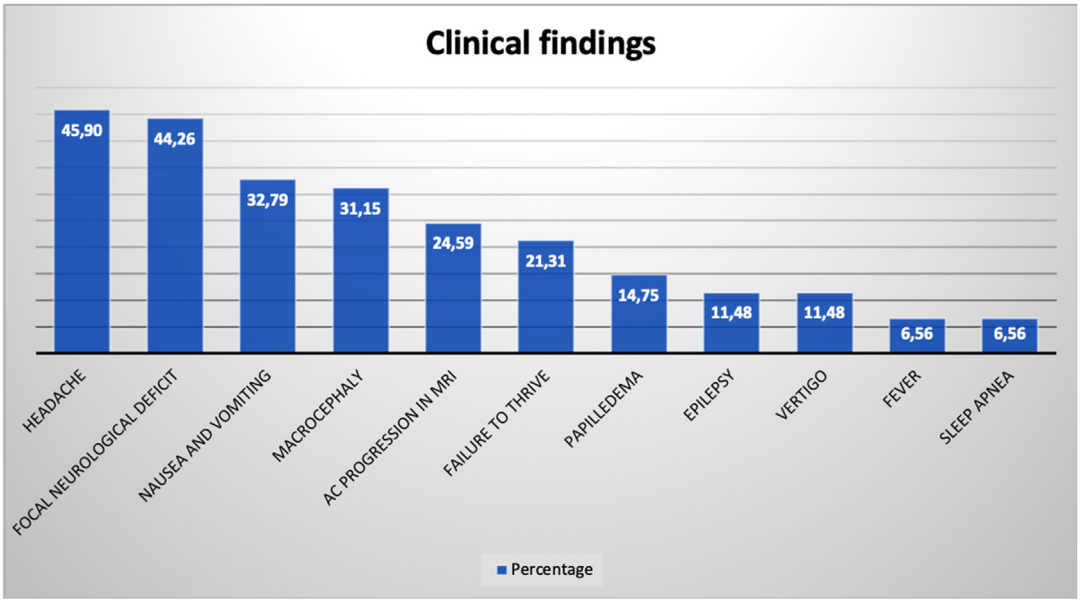

Fig. 2. Clinical findings before surgery.

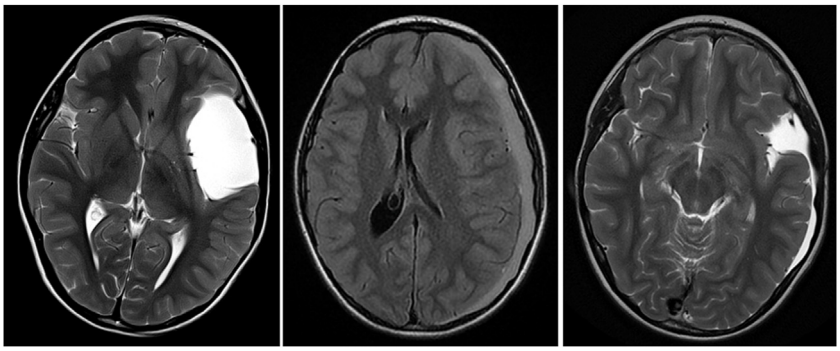

Fig. 3. a: axial T2 MR image showing left-sided perisylvian arachnoid cyst. **b:** axial T2 FLAIR MR image 2 months after surgery and one week after head trauma showing subdural hematoma. **c:** 6 months postoperative axial T2 MR image showing marked decrease in cyst volume with a residual "silent" subdural hygroma.

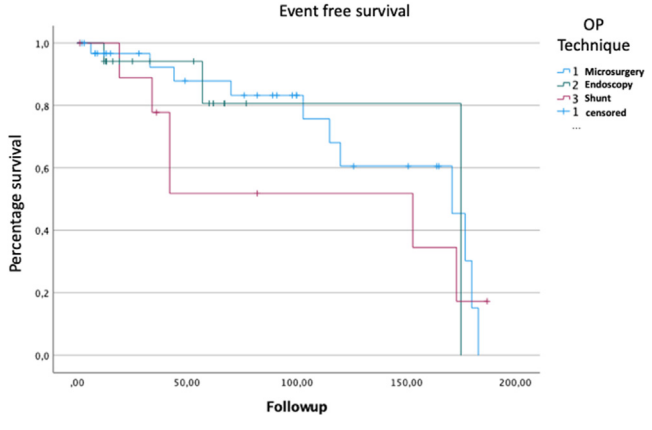

Fig. 4. Kaplan-Meier-Curve defined as "Event free survival" between all three techniques in all locations of ACs.

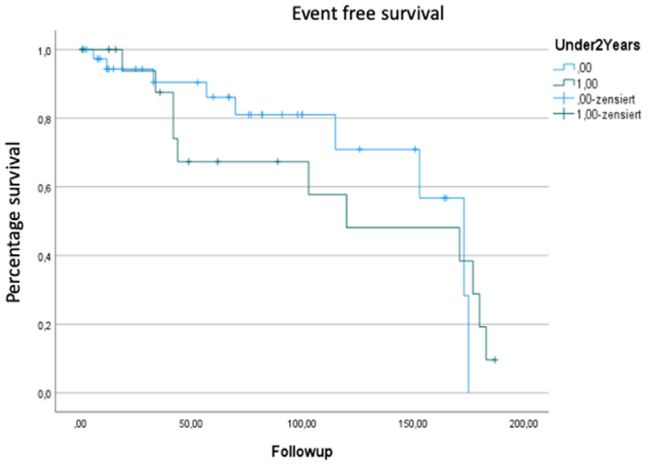

Fig. 5. Kaplan-Meier-Curve defined as "Event free survival" between all three techniques in patients below 2 years of age.

free vs 31/40 patients >2 years, $\mathbf{p = 0.02}$. The need for shunt later after MS or ES was significantly higher in that age group after exclusion of patients with initially implanted CPS; in 6/14 patients <2 years versus 4/37 patients >2 years, $\mathbf{p = 0.02}$. See Fig. 5 and Table 3.

3.6. *Perisylvian ACs subgroup*

Representing the largest number with 31/61 ACs in our cohort, we performed a detailed analysis of the results in that specific location trying

Table 3
Comparison between patients <2 and patients >2 years of age.

	Patients <2 years old $n = 21$	Patients >2 years old $n = 40$	*Significance*
Location	Perisylvian: 6 Suprasellar: 1 Interhemispheric: 10 Retrocerebellar: 3 Cerebellopontine angle: 0 Others: 1	Perisylvian: 25 Suprasellar: 3 Interhemispheric: 2 Retrocerebellar: 2 Cerebellopontine angle: 5 Others: 3	*p=<0.01*
Operative technique (%)	MS 9 (42.9%) ES 5 (23.8%) CPS 7 (33.3%)	MS 24 (60%) ES 13 (32.5%) CPS 3 (7.5%)	***p = 0.04***
Postoperative Hygroma (%)	2 (9.5%)	5 (12.5%)	*NS*
Events at 6 m (%)	5 (23.8%)	6 (15%)	*NS*
Events at 12 m (%)	6 (28.6%)	7 (17.5%)	*NS*
Events at 48 months (%)	11 (52.4%)	9 (22.5%)	***p = 0.02***
Long-Term CPS Implantation (%)	6/14[a] (42.9%)	4/37[a] (10.8%)	***p = 0.02***

[a] After excluding patients with initial CPS-Implantation ($n = 10$).

to find out which operative technique is superior. 31 perisylvian ACs (Galassi type 2 or 3) were operated via MS in 17 patients, ES in 9 patients and 5 CPS were inserted. We noticed that the mean age of patients where CPS were inserted was significantly lower, 2.8 ± 3.4 years in CPS group versus 9 ± 5.2 years in ES group and 8.9 ± 5 years in MS group), **p = 0.03**. Subdural hygroma occurred in 4/17 after MS, 1/9 after ES and in none after CPS, **p = 0.5**. Clinical improvement was noticed in 16/17 patients after MS, 8/9 patients after ES and in all patients of CPS group. Similar results were encountered regarding radiological improvement; 12/17 after MS, 7/9 after ES and 4/5 after CPS. Insertion of CPS was needed in 3/17 patients after MS versus 1/9 patients after ES.

ES demonstrated significantly better event-free survival than MS (8/9 patients versus 12/17 patients, respectively) and CPS (1/5 patients), **p = 0.04**. This benefit was evident in the long term, as seen at the 6-month checkpoint where 4/17 revisions were needed after MS compared to 1/9 revisions after ES and no revisions after CPS. At the 12-month checkpoint, only one more revision was needed following CPS. At the 48-month checkpoint, 4/5 patients in the CPS group required revision surgery, in addition to one more patient in the MS group. Therefore, the endoscopic technique is superior to both MS and CPS for patients with perisylvian ACs in the short and long term. See Fig. 6 And Table 4.

4. Discussion

4.1. Summary of results

To our knowledge, this is the first retrospective cohort study, which directly compares the effectiveness, complication, and revision rates between the available 3 surgical treatment techniques for intracranial ACs in pediatric patients, all treated in one neurosurgical facility. All three techniques (microsurgery, endoscopy and cystoperitoneal shunt) showed postoperative clinical and radiological improvement. Although, the shunt group showed the highest rate of clinical (100%) and radiological (90%) improvement, it showed the highest long-term revision rate (60%). Microsurgery showed the lowest short-term event free survival after 12 months but was superseded by shunt group on long-term basis after 4 years. Thus, endoscopic treatment was proven to be a safe and effective treatment on short and long terms.

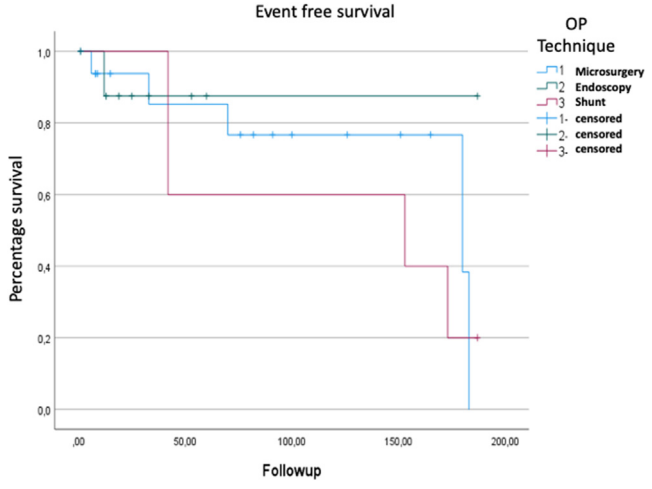

Fig. 6. Kaplan-Meier-Curve defined as "Event free survival" between all three techniques in only perisylvian ACs.

Table 4
Summary of results of perisylvian ACs ($n = 31$).

Operative technique	MS	ES	CPS	*Significance*
Number (%)	17 (54.8%)	9 (29%)	5 (16.2%)	*NS*
Age in years	8.9 ± 5	9 ± 5.2	2.8 ± 3.4	***p = 0.03***
Clinical improvement (%)	16 (94%)	8 (89%)	5 (100%)	*NS*
Radiological improvement (%)	12 (71%)	7 (78%)	4 (80%)	*NS*
Postoperative Hygroma (%)	4 (23.5%)	1 (11%)	0 (0%)	*NS*[a]
Events at 6 m (%)	4 (23.5%)	1 (11%)	0 (0%)	*NS*[a]
Events at 12 m (%)	4 (23.5%)	1 (11%)	1 (20%)	*NS*
Events at 48 months (%)	5 (29.4%)	1 (11%)	4 (80%)	***p = 0.04***
Long-Term CPS Implantation (%)	3 (17.6%)	1 (11%)	–	*NS*

NS: not significant.
[a] MS vs ES.

4.2. Indication for surgery

An individualized treatment strategy should be applied when it comes to selecting patients for surgical treatment. Due to the high incidence of complications 6–20%,[6,11,14,17,17,23] many studies favored conservative approach.[24,25] Most authors agree that symptomatic arachnoid cysts necessitate surgical treatment,[3,6,8,11] still some reserve surgery only for patients with definite symptoms of raised ICP.[25] ACs may also impair cognition and can be responsible for psychiatric disorders as well as neuropsychological defects, which for some could be reversible after surgical decompression.[26–28] In our study, we considered surgery whenever symptoms of increased ICP develop, or signs of neurological deficit were detected. Surgery was in rare cases advocated after neuropsychological testing of children which showed a mild neurocognitive deficit correlated with the cyst location and the possible effect of the local increased intracystic pressure.[29]

4.3. Choice of treatment

Patients below 2 years of age harboring an arachnoid cyst commonly present with macrocephaly. These patients are more likely to require shunts compared to others presenting with other findings, such as seizures or incidental lesions. Accordingly, they are more prone to complications and revisions.[30] In our study, we found that children below 2 years of age have received significantly more CPS for their arachnoid cysts and have significant lower event free survival. Hence, we elaborated the need to shift more towards endoscopy or microsurgery in this age group to avoid the high revision rate and long-term consequences of shunt dependancy.

In addition to other common complications of CSF shunting (infection, obstruction, disconnection, etc.), CPS insertion is also capable of producing shunt dependency,[31,32] with consecutive hazards related to excessive drainage, for example, orthostatic headache, slit cyst syndrome,[32] posterior fossa crowding,[33] craniocerebral disproportion[33–35] and acquired (pseudo) Chiari malformation.[36,37] Hence, most neurosurgeons nowadays try to avoid placing CPS and to resort to using microsurgical techniques or neuroendoscopic procedures for cyst fenestration instead. We encountered these changes in one of our patients with CPS and we could stop the cascade of complications through a valve revision that has been set to the highest pressure planning to go for endoscopic fenestration in the near future after restoring adequate cyst size feasible for fenestration.

Our study revealed a shift in treatment preference over time. Initially, CPS insertion was favored due to its relative ease and low risk of surgical complications, particularly in small children below 2 years of age. However, as long-term complications and revisions associated with CPS insertion became more apparent (60%), MS became more prevalent in an effort to reduce the need for CPS insertion. More recently, we observed a significant shift towards ES, which provided effective treatment with better event-free survival in both the short and long terms.

4.5. Clinical versus radiological improvement

There is lack of evidence whether there is a relationship between postoperative size reduction of the cyst and clinical improvement.[25,28] Amelot et al[28] assumed that the best markers of a successful treatment were the improvement of clinical and/or cognitive signs and the presence of a stream that is visible at the cystostomy level on MRI 3D T2-weighted sequences. In our study, we encountered clinical improvement in 95% of patients (58/61) following surgery and radiological improvement in 83.6% (51/61 patients) which proves also that clinical improvement can occur without significant reduction of cyst size morphologically as we believe that the elevated pressure is the main drive for symptoms but not the size itself. Nevertheless, we expect a consequent decrease in size after pressure release but not in all cases depending on the initial size.

4.6. Perisylvian arachnoid cysts

Perisylvian ACs are known to have the highest complication rate in comparison to other locations of ACs.[38] Chen et al[39] tried to figure out the efficacy and safety of the 3 surgical options (ES, MS, and CPS) for treatment of perisylvian ACs, they performed a meta-analysis including 18 studies with 474 patients, half of them operated endoscopically, they concluded that all 3 surgical methods are effective but considering safety, neuroendoscopic fenestration may be the best initial procedure. This has also been proved in our study as we found that the event free survival in our subgroup of 31 perisylvian ACs also favors ES (88.9%) rather than MS (70.6%) or CPS (20%).

4.7. Limitations of the study

The study has some limitations due to its retrospective and monocenter nature, as well as the inequality of the study groups in terms of follow-up length and the number of surgeons involved. A randomized, blinded allocation of patients to the study groups would have increased the validity of the results but was not feasible due to the personal preferences of the surgeons over the long study period. Despite these limitations, it is worth noting that this is the first comparative study on the treatment of pediatric intracranial ACs using MS, ES, and CPS techniques, conducted in a single neurosurgical center.

It is challenging to determine the most effective surgical option for arachnoid cysts when considering all of their different locations, as outcomes may differ significantly, and the surgeon's experience plays a crucial role in decision-making. Nonetheless, this selection bias was at least in the best interest of the patients, as the choice of treatment method was based on the surgeon's most experienced surgical technique and a desire to minimize harm. The difference in follow-up duration among the three modalities may introduce bias, as longer follow-up intervals are more likely to reveal complications.

Providing information on the total number of patients with arachnoid cysts who were not operated due to the absence of symptoms would have given us a better understanding of the overall patient population and the criteria for surgery. However, this data was not available as these patients were only referred to us by pediatricians once they exhibited symptoms. Consequently, there may have been children who were not included in our study database as they did not receive a neurosurgical consultation.

5. Conclusion

Endoscopic fenestration is a safe and efficient technique which is being widely used nowadays as a treatment of symptomatic intracranial arachnoid cyst. Microsurgical fenestration is efficient but with a lower event free survival in comparison to endoscopic technique. Although insertion of cystoperitoneal shunt carries the least surgical risks but during follow-up shows the highest revision rate. On long terms, CSF volume depletion, together with the resultant reduction in cranial capacity, leads to a cascade of events that are responsible for shunt dependency and craniocerebral disproportion. Hence, nowadays most neurosurgeons try to avoid placing cystoperitoneal shunts and resort to using microsurgical techniques or neuroendoscopic procedures for cyst fenestration instead.

Authors' contributions

AeD, MI, FP wrote the main manuscript text.
All authors reviewed the final version of the manuscript.

Ethical approval

The study was approved by our ethic committee (S-084/2022).

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

List of abbreviations

AC	Arachnoid cyst
CISS	Constructive interference in steady state
CPS	Cystoperitoneal shunt
CSF	Cerebrospinal fluid
ES	Endoscopic surgery
ICP	Intracranial pressure
MRI	Magnetic resonance imaging
MS	Microsurgery

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