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Characteristic and management of pediatric arachnoid cysts: A case series

Abdulfettah Tümtürk [1], Nimetullah Alper Durmuş [2], Süreyya Burcu Görkem [3], Murat Çiftçi [1], Mehmet Canpolat [4], Hüseyin Per [4]

[1] Department of Neurosurgery, Acıbadem Hospital, Kayseri, Turkey
[2] Department of Neurosurgery, Kayseri City Training and Research Hospital, Kayseri, Turkey
[3] Department of Radiology, Adana City Hospital, Adana, Turkey
[4] Department of Pediatric Neurology, Erciyes University School of Medicine, Kayseri, Turkey

ORCID ID of the author(s)

AT: 0000-0001-5883-9819
NAD: 0000-0002-7581-790X
SBG: 0000-0001-8949-6476
MÇ: 0000-0003-2780-1804
MC: 0000-0002-2197-8433
HP: 0000-0001-9904-6479

Corresponding Author
Nimetullah Alper Durmuş
Specialist in Neurosurgery, Kayseri City Training and Research Hospital, Kayseri, Turkey
E-mail: nimetullahalper@hotmail.com

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Ethics Committee approval was taken from the Erciyes University, Medical Faculty, Clinical Research Ethics Committee (date: 1/08/2012, decision number: 541).
All procedures in this study involving human participants were performed in accordance with the 1964 Helsinki Declaration and its later amendments.

Conflict of Interest
No conflict of interest was declared by the authors.

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Abstract

Background/Aim: Arachnoid cysts (ACs) are the collection of fluid, which is similar in composition to cerebrospinal fluid, within the congenitally duplicated arachnoid membrane. ACs are clinically silent lesions, but sometimes they can manifest themselves with headache, convulsion, focal neurological deficits, cognitive decline, torticollis, and macrocephaly. Although the appropriate surgical approach is less clear, there is a consensus on the need for surgical treatment in symptomatic ACs. This study aims to reveal the advantages and disadvantages of cystoperitoneal shunt and microsurgical fenestration techniques.

Methods: One hundred ninety-one consecutive patients from 1 month to 15 years old with AC were evaluated for suitability. Sixteen patients who underwent surgery were included in the study. Medical records of the patients with AC managed at our institutions were retrospectively collected and analyzed. Sixteen of 191 patients underwent surgical treatment via craniotomy for microsurgical cyst fenestration (CF) and cysto-peritoneal shunting (CS). CF was performed with partial cyst wall excision in all patients.

Results: Seizure was the most common presentation in the patients, followed by headache and trauma, respectively. In our series the most common indications of the surgery were increased intracranial pressure (IICP). CF was performed in nine patients, and CS was performed in seven as the primary treatment. CS-related complications, such as infection (n = 2), dysfunction (n = 2) and intraabdominal abscess (n = 1) were the most commonly observed complications. No infections were observed after CF, but subdural hematoma was observed in one child.

Conclusions: Although the most common symptoms in cases with arachnoid cysts are headache, rare symptoms, such as torticollis, may exist. Due to this, clinicians should reveal the cyst-complaint relationship first. Given the increased complications of CS, in addition to shunt independency and being free from shunt-related complications, the microsurgical CF with wide excision of the membranes seems to be the more preferable surgical option.

Keywords: Arachnoid cysts, Conservative management, Craniotomy, Cysto-peritoneal shunting, Microsurgical cyst fenestration

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Introduction

Arachnoid cysts (ACs) are the collection of fluid, which is similar in composition to cerebrospinal fluid within the arachnoid membrane. It is thought to result from splitting or duplication of the primitive arachnoid membrane in early embryonal life [1, 2]. These benign congenital cystic lesions occur in 2.6% of children [3, 4]. The incidence of ACs is reported to be about 1% of intracranial space-occupying lesions. Males are involved in more than two-thirds of the cases and there is an increased incidence on the left side [5].

ACs most frequently arise in the supratentorial space, especially in the middle cranial fossa, followed by the convexity and suprasellar regions. Only 10% of ACs arise in the posterior fossa [1, 4].

In general, ACs are clinically silent lesions. Sometimes they can manifest themselves with clinical symptoms and signs, such as headache, convulsion, focal neurological deficit, and cognitive decline like intellectual disabilities and torticollis [6-10]. In addition to these, thinning of the adjacent bone and macrocephaly may also be seen. These clinical manifestations depend on the location, size and mass effect of ACs [8-12].

Although the appropriate surgical approach is less clear, there is a consensus on the need for surgical treatment in symptomatic ACs [12-15]. In symptomatic ACs, the treatment options vary from open craniotomy and endoscopic approaches to cystoperitoneal shunts and stereotactic aspirations [12, 14-17]. In asymptomatic ACs, conservative management has been proposed and prophylactic surgery is generally not recommended. For these patients, a close radiological and clinical follow-up is of great importance [2, 4, 18].

This study aims to reveal the advantages and disadvantages of the cystoperitoneal shunt and microsurgical fenestration techniques.

Materials and methods

PatieIn this study, we evaluated the clinic, radiologic, demographic findings, and therapeutic approaches and prognoses of children with arachnoid cysts. We have aimed to evaluate the clinical and radiological course of the disease as well as the impact of clinical symptoms and radiologic findings of children with ACs to best determine the treatment modalities. We have also compared the results of two different surgical approaches in the enrolled children. One hundred ninety-one consecutive patients with AC diagnosed and treated in the Department of Neurosurgery, Erciyes University Medical Faculty from August 2012 to June 2014 were included in the study. Medical records of all pediatric patients with ACs who underwent surgical treatment and were conservatively managed at our institution were analyzed.

The data collected included age, gender, neurologic symptoms and signs, associated abnormalities, psychomotor status, surgical intervention, complications, electroencephalography findings, radiologic findings, and follow-up. Patients with secondary ACs due to infection or trauma were excluded from the study.

All patients were evaluated preoperatively using computed tomography (CT) and/or magnetic resonance imaging (MRI) scans. Postoperatively all patients had regular follow-up clinical and radiological evaluations. The mean follow-up period was 61 months (range, 14-136 months).

Informed consent forms from the patients' parents were obtained for all patients.

Cranial CT

All of the patients underwent a low dose non-enhanced multi-detector cranial CT (Toshiba Aquilion One 320 slice CT, Toshiba Medical Systems, Otawara, Japan) (120 kV, 10mA). None of the patients were sedated.

Cranial MRI

Patients younger than age five were sedated with midazolam (intranasal 0.1-0.2 mg/kg, Dormicum, Roche, Istanbul, Turkey) for brain MRIs. Patients five years old and older underwent brain MRIs without sedation in the supine position. A total of 191 patients underwent a routine pediatric brain MRI examination (1.5 T Philips Intera, Philips Medical System, Best, The Netherlands and 1.5 T Siemens Aera, Siemens Medical Solutions, Erlangen, Germany) in our institution. Our pediatric brain MRI protocol included T1-W axial-sagittal, T2-W axial-coronal, T2-W FLAIR axial, DWI and ADC images. The slice thickness varied from 4 mm to 5 mm.

Sixteen (9 males and 7 females aged from 1 month to 15 years old) out of 191 patients underwent surgical treatment via craniotomy for microsurgical cyst fenestration (CF) and cystoperitoneal shunting (CS). CF was performed with partial cyst wall excision in all the patients. We performed CS in one patient whose parents refused the CF operation. Three of the seven patients in the CS group had been operated via CS in other centers before admission to our department. In all of the 16 patients in surgical group sulcal obliteration, shifting of adjacent vascular structures, adjacent cortex depression or ventricle effacement or enlargement, and midline shift were observed.

We performed CF in nine patients and CS in seven as a primary treatment. In three of the seven cases who underwent CS, we performed CF due to shunt-related complications (two instances of shunt revision, two cases of shunt dysfunction, one shunt infection with intraabdominal abscess, and one case of meningitis).

Statistical analysis

Statistical analyses were performed with R [R project (2014). R (Version 3.1.1), computer software, retrieved from https://www. r-project.org). To summarize the data obtained from the study, descriptive statistical data are presented as mean (standard deviation) and median (range) for continuous variables based on data distribution. Categorical variables are summarized as count and percentage.

Results

The median (min-max) age of the patients (121 male and 70 female) was 5.0 years (ranging from 1 month to 18 years) for both genders. Seizure was the most commonly presented symptom and indication for MRI in the patients, and headache and trauma followed respectively. In this study of 191 children under 18 years of age with AC, indications for MRI are given in Table 1.

Figure 1 (a, b, c, d, e, f): 17-month-old boy with torticollis: A giant arachnoid cyst with right cerebellar hypoplasia is shown on T1-W (a), T2-W (b) axial and T2-W (c) coronal images. Cyst size reduction and no brainstem compression are seen on the postoperative late control T1-W (d), T2-W (e) axial and T2-W (f) coronal images of the case.

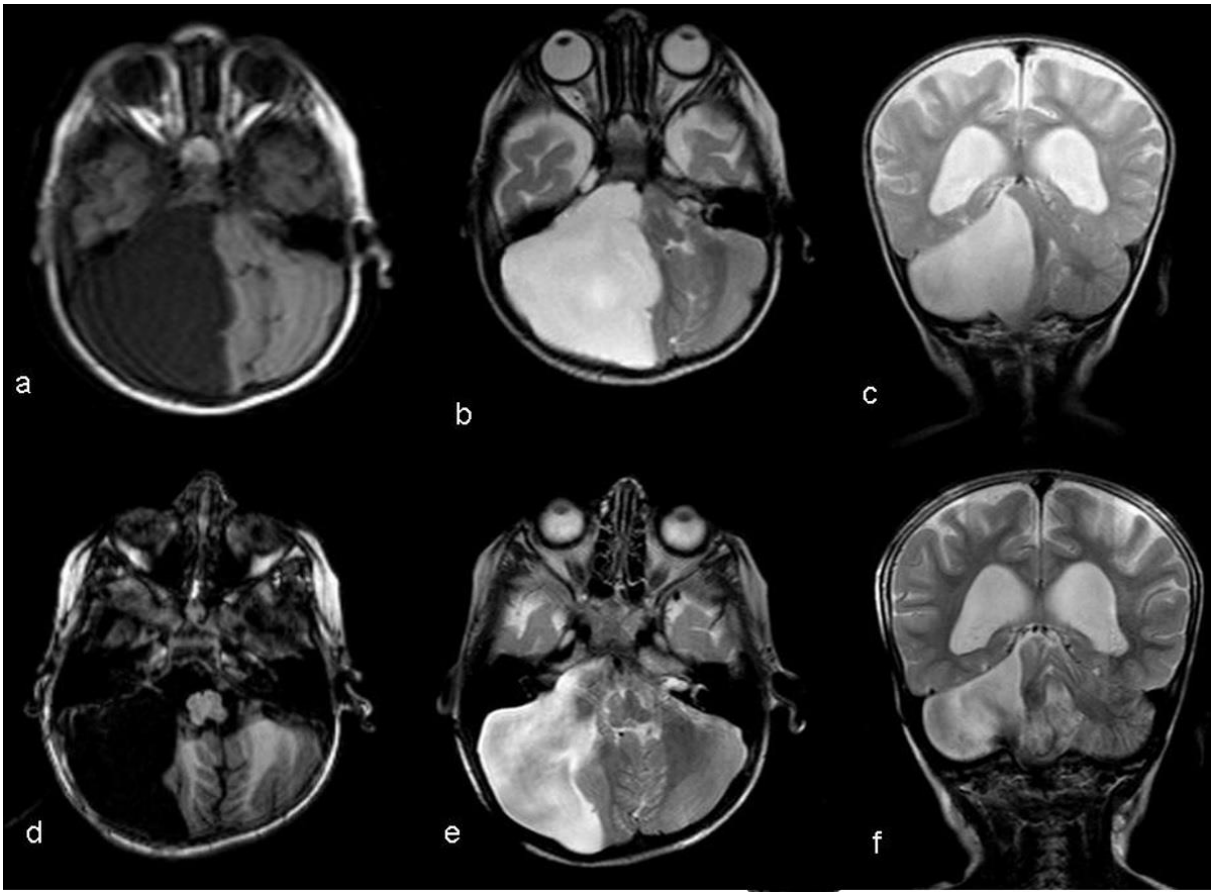

Figure 2 (a, b, c, d): 11-month-old boy: A giant arachnoid cyst at left frontotemporal region, which compresses the left lateral ventricle and causes a midline shift is demonstrated on T1-W (a) sagittal and T2-W (b) coronal images (arrows). After the CF, the cyst regressed totally but still remains at left temporal region on T1-W (c) sagittal and T2-W (d) coronal images (arrows).

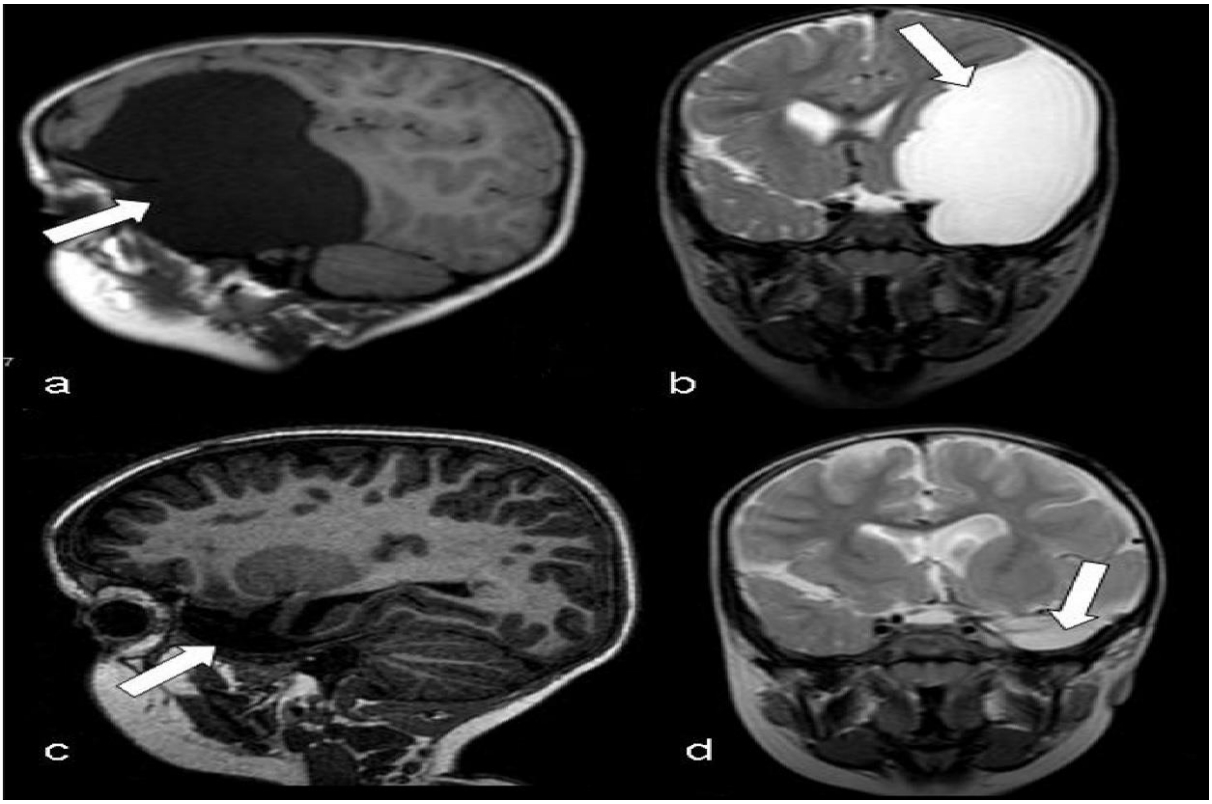

Table 1: Initial presentation symptoms of 191 children with arachnoid cysts

Indication	No	Case %
Concern for seizure	74	38.9
Headache	34	18.0
Trauma	16	8.0
Headache with vomiting and ataxia	11	6.0
Cognitive dysfunction or developmental delay	12	6.2
Signs related to the eye	12	6.2
Pitosis 1		
Strabismus 9		
Nistagmus 2		
Gait Disturbance	9	5.0
Macrocephaly	8	4.1
Abnormal and involuntary movement	5	3.0
Vertigo	4	2.1
Pituitary/endocrine issues	1	0.05
Cutaneous lesion	2	1.0
Torticollis	1	0.05
Other (detected in fetal MRI)	2	1.0

One hundred thirty-four of the ACs were supratentorial (70.2%) and 57 were infratentorial (29.8%). The left hemisphere was more often affected (n = 80). Lesion locations are given in Table 2. During the follow-up period, even though there were no clinical complaints and neurological symptoms in 175 of 191 cases with ACs, spontaneous resolution in AC was observed in 6 patients. However, in 17 cases, there was a slight growth in their cysts, but no clinical or neurological findings were observed.

Table 2: Location of arachnoid cysts in 191 pediatric patients with arachnoid cysts

Lesion location	Total No.	% Cases
Anterior fossa	12	6.3
Middle fossa	96	50.3
Left temporal 61		
Right temporal 29		
Bilateral 6		
Posterior fossa	52	27.3
Left cerebellar 15		
Right cerebellar 25		
Cisterna magna 12		
Cerebellopontine angle	4	2.1
Convexity	1	0.05
Ventricular region	11	6.0
Bilateral lateral ventricle 2		
Right lateral ventricle. 4		
Left lateral ventricle 1		
3rd ventricle 3		
4th ventricle 1		
Basal cisterns	8	4.2
Sellar/suprasellar region	1	0.05
Interhemispheric region	6	3.2
Total	191	

Anterior fossa → right frontotemporoparietal n:1, frontotemporal n:4 (2 right, 2 left), right frontoparietal n:1, frontal n:6 (5 right, 1 left)

During the study period, only 8.4% of the patients with AC (16/191) underwent surgery via microsurgical CF and/or a CS procedure. This included 9 males and 7 females with a mean age at the time of surgery of 57.8 months (ranging from 1 month to 15 years). Of the 16 ACs, 12 were in the supratentorial and 4 were in the infratentorial location.

Table 3: Clinical and imaging details and surgical indications for 16 pediatric patients with arachnoid cyst who underwent surgical treatment

Case No	Age/Sex	Sign/Symptom	Cyst Location	Cyst Size (mm) Initial	Surgical indications	Surgical Procedure	Cyst Size (mm) Postoperative	Complication CS / CF	Clinical Improvement
1	17 m/M	Torticollis	PF	64×74×55	Torticollis	CF	41×52×60	none	No Torticollis
2	31 m/M	Convulsion	Right LV	40×30×35	Hydrocephaly	CS, CF	30×18×27	Shunt Dysfunction / none	Normalized skull skull growth rate
3	122 m/F	Convulsion, Ataxia	PF	68×62×41	Brainstem Compression, MEC	CF	68×52×43	none	No Ataxia
4	94 m/M	Headache, self- harm	Interhemispheric	78×42×69	MEC	CF	70×20×50	/CSDH	No headaches
5	38 m/F	Macrocephaly	PF, Occipital	132×110×143	Hydrocephalus	CS	94×94×124	SDHy	Normalized skull skull growth rate
6	1 m/M	Macrocephaly	Left LV	57×65×63	Hydrocephalus	CS	34×58×41	SDHy	Normalized skull skull growth rate
7	56 m/M	Strabismus	3rd ventricle	37×42×42	Strabismus, Hydrocephalus	CF	31×21×52	none	Normalized skull skull growth rate No Strabismus
8	17 m/F	Strabismus	3rd ventricle, prepontine–interpedincular cisterna	31×34×25	Strabismus, Hydrocephalus	CF	31×21×32	none	Normalized skull skull growth rate No Strabismus
9	156 m/F	Polydips ia, gait disturbance, vertigo,	Prepontine cisterna, suprasellar	32×30×33	Hydrocephalus, Brainstem Compression	CS	20×26×15	Meningitis / none	No Polydipsia and gait disturbance
10	7 m/M	Vomiting, irritability	interhemispheric	60×50×45	Hydrocephalus	CS	30×20×33	none	Normalized skull skull growth rate No Vomiting
11	21 m/M	Macrocephaly, vomiting	Left Frontotemporal	70×58×98	MEC, macrocephaly	CF	33×36×17	none	Normalized skull skull growth rate No Vomiting
12	8 m/F	Macrocephaly	Right LV	79×65×65	Hydrocephalus, macrocephaly	CS, CF	71×30×64	Shunt dysfunction / none	Normalized skull skull growth rate
13	82 m/F	Headache, Fainting	PF	52×42×46	MEC	CF	29×31×28	none	No headaches and fainting
14	84 m/M	Headache, blurred vision, vomiting, papilledema	Right Frontotemporo-parietal	76×49×43	MEC	CF	38×21×12	none	Symptoms of IICP improved
15	180 m/F	Headache, vomiting, papilledema, confusion	Ambient cisterna	35×31×19	MEC, Hydrocephalus	CS, CF	10×14×11	Shunt infection, intra abdominal abscess / none	No headaches and fainting
16	11 m/M	Headache, vomiting	Left Frontotemporal	85×61×59	MEC	CF	26×20×16	none	No headaches and vomiting

m: Month, M: Male, F: Female, PF: Posterior fossa, LV: Lateral Ventricle, CF: Cyst fenestration, CS: Cyst shunting, CSDH: chronic subdural hematoma, SDHy: subdural hygroma, MEC: Mass effect of the cyst

In two patients less than one year of age who were administered CF, we did not come across any complications. The result was both clinically and radiologically a success. A 17-month-old boy with torticollis and a right cerebellar arachnoid cyst (Figure 1) and an 11-month-old boy with a giant arachnoid cyst at the left frontotemporal region (Figure 2) were given as sample cases.

There were no deaths in our series. No infections were observed after CF, but we did meet with subdural hematoma in one case, which was treated surgically. Shunt-related complications, such as infection and dysfunction, were the most common complications in the included patients. Additionally, we found subdural hygroma in two patients treated with CS. These were managed conservatively.

In our series, the most common indications of the surgery were IICP due to mass effect of the cyst (n = 6) or hydrocephalus (n = 9) that was related to AC. The other indications included strabismus (n = 2), brain stem compression (n = 2), macrocephaly (n = 2) and torticollis (n = 1), respectively. No patients were operated on with an indication of cyst size alone. Clinical, surgical, and imaging details of 16 pediatric patients with AC who underwent surgical treatment are given in Table 3.

Discussion

The most common locations of ACs are reported to be middle cranial fossae (80%) and posterior cranial fossae (10%), whereas convexity lesions constitute only 5% of all [1, 13]. In our series, 134 of the ACs were supratentorial (70.2%), and 57 were infratentorial (29.8%).

ACs are CSF collections usually incidentally found on MRI/CT and usually do not enlarge. Even so, they rarely disappear spontaneously. In our series, spontaneous resolution in AC was observed in six patients. Most ACs are static, typically clinically silent but have the potential to enlarge and surgical intervention is absolutely required when sufficient cyst enlargement occurs. Asymptomatic and incidental cysts do not require treatment or should not be treated surgically, but they should be followed up regularly. All authors agree that symptomatic ACs, which are presented with IICP, intractable seizures, and focal neurological deficits should be treated surgically [14, 18].

In our series the most common indications of the surgery were IICP due to mass effect of the cyst or hydrocephalus that was related to AC. The other indications of the surgery included strabismus, brain stem compression, and torticollis, respectively.

Seizure may not always be an indication for surgical intervention when originated from the dysmorphic cortex beneath the seizure or originated from a different region of the brain [16]. Although the most common presentation in 191 patients was seizure, there were only two patients with seizure complaints in the surgical group, whose indication was not related to the seizure but hydrocephalus and brain stem compression due to ACs. No patients were operated on due to intractable seizures in our series.

Although neurological symptoms exist, clinicians should exactly correlate nonspecific symptoms with AC, because ACs can present themselves with uncommon symptoms, such as torticollis in a 17-month-old boy in our series. This patient completely recovered from torticollis after the surgery. In the surgical group of our study, all the complaints of the patients were examined regardless of whether they were correlated with AC. Most of the complaints were related to IICP due to hydrocephalus and/or mass effects of the cyst.

The choice of the most appropriate surgical approach to the treatment of pediatric arachnoid cysts remains widely debated [12-14]. The treatment options vary from microsurgical resection of the cyst wall, fenestration of the cyst to the ventricle or cisternal space with open craniotomy or endoscopic approaches to CS [19, 20]. However, each treatment modality has some advantages and disadvantages. Nevertheless, CS placement seems easier, and it is possible to achieve high rates of cyst elimination with this treatment. However, CS carries the additional risk of shunt-dependent complications, such as shunt infection, shunt failure, unexpected hemorrhage, and over drainage. Additionally shunt-related complications may occur at any time in life [5].

Both of the open craniotomy and endoscopic procedures for cyst excision or fenestration have the advantage of leaving the patient shunt independent [11, 13, 15]. However, surgical techniques require a slow learning process and the learning curve should be completed before using them as the first line of treatment [16]. In addition, it is difficult to fenestrate the cyst into a CSF cistern or ventricle and to perform cyst wall resectioning using this technique. Overall, the rate of radiological resolution of the cyst has been reported to be lower in endoscopic approaches.

Another important disadvantage of this procedure is controlling unexpected profuse bleeding due to difficulties in instrument use and insufficiency in view. These problems are not encountered in microsurgery [4, 17]. In addition, the location of the cyst may impose significant risk to surrounding structures, as is the case with suprasellar or posterior fossa cysts [4]. CF procedure is not always effective, and cyst recurrence has been reported [13, 20]. It has some complications including meningitis, hemiparesis, subdural hematomas, seizure, and even death [4, 19].

Because surgical therapy is more effective in children, it is recommended that ACs that show poor communication between the cyst and the subarachnoid spaces should be treated as early as possible with regard to reversible brain growth in childhood [11].

The results in patients under one year of age who were administered CS were radiologically and clinically successful in this series. Nevertheless, in an eight-month-old patient, due to recurrent shunt problems, we had to perform CF. In two patients less than one year of age who were administered CF, we did not come across any complications. The results were both clinically and radiologically successful. Even though a relatively high surgical failure rate was previously reported in pediatric patients under one year of age [18], due to the success of the procedure in this age category, we are of the opinion that in patients under one year of age, CF is possible to be performed.

Both subdural hygromas and subdural hematomas have been previously reported after the treatment of ACs in children,

whether microsurgical or endoscopic [17, 20]. In the patients treated with CS, we observed shunt infection in two patients, shunt dysfunction in two, and subdural hygroma in another two. In only one of the cases treated with CF did we observe acute subdural hematoma. No infection was experienced. The rate of complications in our series, especially infection in the CF group, was significantly less than in the CS group.

Late follow-up imaging data showed nearly complete resolution of the cyst in 12.5% of the 16 patients. In addition, there was a significant reduction in size in 50% of cases, while 37.5% of the cysts slightly decreased. In one patient, although the cyst size was decreased in early control, it was found to be in similar dimensions in late control. We concluded that the reason for this might have been the insufficient fenestration of the cyst or the closure of fenestration. Even the radiological improvement was not observed or the cyst did not change in size. Clinical symptoms of IICP in the cases, such as headache (n = 4) and vomiting (n = 4), were improved and normalized skull growth rate was obtained in eight patients. Other improved symptoms included ataxia (n = 1), torticollis (n = 1), strabismus (n = 2), gait disturbance (n = 1), polydipsia (n = 1) and fainting (n = 2). There was no exact correlation between radiological and clinical improvements.

Although it has been previously reported that patients who show radiological improvement do not always demonstrate clinical improvement [18], the patients who showed radiological improvement always demonstrated a corresponding improvement of clinical symptoms in the presented patients. Although we observed a recurrence of the size after surgery, we speculate that these procedures are clinically effective, since the symptoms and complaints of the patients clearly improved.

However, a weakness of this study is that it consisted of a small number of surgical cases. Due to this, generalizability of the results is difficult.

Conclusion

Although the most common symptoms in cases with arachnoid cysts are headache, rare symptoms, such as torticollis, may exist. Due to this, clinicians should reveal cyst-complaint relationship first.

This study aimed to reveal the advantages and disadvantages of cystoperitoneal shunt and microsurgical fenestration techniques.

Cystoperitoneal shunting is likely to be a technically easier surgical treatment modality in children. However, lifelong shunt dependency and a lifelong risk of shunt-related complications are the serious disadvantages of this treatment modality. The microsurgical fenestration into more than one space of the cyst with wide excision of the membranes is a preferable option for surgical treatment.

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