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ORIGINAL ARTICLE

Suprasellar arachnoid cysts in adults: clinical presentations, radiological features, and treatment outcomes

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Abstract

A tendency for suprasellar arachnoid cysts (SACs) to occur in young children is known. Data of adult SACs were rare in previous reports. The aim of this study is to discuss their clinical presentations, radiological features, and treatment outcomes based on 23 adult patients who underwent endoscopic fenestration in our hospital between January 2003 and December 2018. Preoperative cyst volume ranged from 12.3 to 72.5 cm^3 (mean 39.8 ± 19.8). Endocrine disorders occurred in 7 (30.4%) patients. Hydrocephalus was observed in 20 patients. In the patients with hydrocephalus, the mean preoperative Evans' index (EI) (%) and frontooccipital horn ratio (FOHR) (%) were 44.8 (ranged 32.2–63.4) and 49.6 (ranged 36.7–59.8), respectively. A bivariate correlation showed significant positive association between preoperative cyst volume and preoperative EI or FOHR (Pearson correlation, $r = 0.607$, $p = 0.005$; $r = 0.583$, $p = 0.007$). The slit-valve phenomenon was observed in 13 (56.5%) patients. Pale/tenacious cyst walls were observed in 12 (52.2%) patients. Postoperatively, all patients achieved the improvement in clinical symptoms and a decrease in cyst size. The mean decreases in cyst volume, EI, and FOHR were 64.7%, 7.89%, and 5.8%, respectively. A bivariate correlation indicated the irrelevance between the postoperative cyst volume and postoperative EI or FOHR (Pearson correlation: $r = 0.37$, $p = 0.11$; $r = 0.43$, $p = 0.054$). These results reveal that there are a few differences in several aspects between adult patients and child patients. The severity of hydrocephalus is correlated with cyst size in adult patients. Additionally, the excellent outcomes in adult SACs can be obtained by endoscopic fenestration.

Keywords Arachnoid cysts · Adults · Suprasellar · Endoscopic surgery · Hydrocephalus

Suprasellar arachnoid cysts (SACs) are nontumorous and congenital cerebral-spinal fluid (CSF) collections which developed from the abnormality of Liliequist' membrane or interpeduncular cistern [1]. They represent between 8 and 15% of all arachnoid cysts that required treatment [2–4]. The pathogenesis of SAC is unclear, and various explanations have been given to account for this expansion, including a "ball valve" action, an osmotic gradient, slit-valve mechanism, and transudation from choroid plexus tissue remnants or ectopic glial cells [5]. Of them, the slit-valve mechanism is widely accepted [6–8].

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SACs are often said to be rare and can occur at any age, but with a tendency to occur in young children and a male predominance [9–12]. Reviewing previous literatures, the reports on child patients were abundant; however, to date, there were no reports to describe the characteristics of adult SACs. Therefore, the information is void regarding on clinical and radiological features, endocrine evaluations, and the long-term prognosis of adult SACs. The aim of the present study is to discuss these issues based on 23 adult patients with long-term follow-up.

Methods

Patient characteristics and radiological measurements

From January 2003 to June 2018, a total of 254 cases of SACs were treated by using endoscope in our hospital, of which 23 (9.1%) cases of adult SACs were identified and included in

this study. Their clinical charts, surgical records, and videos were reviewed. Radiological data were obtained from picture archiving and communications systems. Three patients with previously inserted ventriculoperitoneal (VP) shunts were admitted for the recurrence of clinical symptoms. The intervals between VP shunt and recurrence were approximately 4, 6, and 16 years. All patients underwent magnetic resonance imaging (MRI) scans in outpatient setting as the initial diagnostic procedure. The cyst volume was measured using the image analysis software ITK-snap 3.8.0 [13]. Evans' index (EI) and frontooccipital horn ratio (FOHR) were used to evaluate the severity of hydrocephalus. The maximal frontal horn width (L_{MFHW}), the maximal occipital horn width (L_{MOHW}), and the maximal transverse inner diameter (L_{MTID}) of the skull at the same level were measured on the preoperative and postoperative (at 1-year follow-up) T2 image on an axial section. EI less than 30% represents no hydrocephalus [14]. All measurements were performed separately by 3 radiologists, and their average measurement results were adopted. Evan's index was equaled to L_{MFHW}/L_{MTID} and FOHR was calculated as $(L_{MFRW} + L_{MOHW})/2\ L_{MTID}$. The slit-valve phenomenon in the mesencephalic leaf of Liliequist was documented as present or absent. The characteristics of cyst walls were described as transparent/thin and pale/tenacious.

Surgical procedures

All endoscopic fenestrations were performed with the patients in the neutral supine position with the head elevated about 20°. An electromagnetic neuronavigational system (Medtronic S7) was used in the patients without hydrocephalus. For the patients with prior VP shunt, the endoscopic fenestration was performed through contralateral ventricle. The optimal bur hole was located at about 3 cm lateral to the midline and about 1 cm anterior to the coronal suture. Endoscopic trajectory was obtained with the help of preoperative MRI to fenestrate both the apical and/or basal cyst membranes with minimal anteroposterior manipulation. After identifying anatomic landmarks of lateral ventricle, fenestration of the apical cyst membrane was performed to complete the ventriculocystostomy (VC) procedure. After VC, the endoscope was advanced into the cyst, which allowed a direct visualization of cisternal contents and the slit-valve phenomenon. If there was a slit-valve, the fenestration of the basal cyst membrane was performed by enlarging the slit-valve using scissors to achieve communication between the cyst and basal cistern. If there was no slit-valve, a fenestration was performed at the avascular site of the basal cyst membrane by using scissors between the cranial nerves and basilar artery.

Follow-up and statistical analysis

Patients were required to perform the periodic clinical and radiological evaluations. The mean duration of follow-up was 52.1 months (ranged 17–85 months). The patients without hydrocephalus were excluded at EI and FOHR measurements. Pearson correlation was used to evaluate the relationship between cyst volume and EI and FOHR. All statistical analyses were performed with SPSS software (version 13.0; IBM Corporation, Armonk, NY, USA).

Results

A total of 23 patients were enrolled in the present study, including 11 (47.8%) male patients and 12 (52.2%) female patients. The mean age was 34.9 years (ranged 18–64). SACs without hydrocephalus were observed in 3 patients (Figs. 1a and 2a, b). Table 1 summarizes the data of all patients. The most common clinical presentations were the symptoms related to increased intracranial pressure (73.9%) and visual compromise (69.6%) including impairment of visual acuity and visual field, followed by endocrine malfunction (30.4%). Hypogonadism occurred in 6 patients, microplasia in 1 patient. Preoperative cyst volume ranged from 12.3 to 72.5 cm^3 (mean 39.8 ± 19.8; median 44.5, interquartile range (IQR) 21.5–57.8). Preoperative EI and FOHR ranged from 32.2 to 63.4% (mean 44.8 ± 8.4) and 36.7 to 59.8% (mean49.6 ± 6.5), respectively. A bivariate correlation showed significant positive association between preoperative cyst volume and preoperative EI or FOHR (Pearson correlation, $r = 0.607$, $P = 0.005$; $r = 0.583$, $P = 0.007$) (Fig. 3).

All patients underwent endoscopic fenestration after admission: VC in 3 patients, ventriculocystocisternostomy (VCC) in 20 patients. In the 3 patients with a prior shunt treatment (Fig. 1a–c), the shunt catheters were ligated with silk thread at clavicle level after endoscopic fenestration to judge shunt dependence. Only one patient had shunt apparatus removed and was stable during the follow-up. At the latest follow-up, no patient required further surgical treatment.

Intraoperatively, the slit-valve phenomenon which was around basilar artery was observed in 13 (56.5%) patients. Cyst wall presented transparent appearance and thin texture in 11 (47.8%) patients, whereas characterized paleness and tenaciousness in other 12 patients.

Postoperatively, all patients achieved the improvement in clinical symptoms and a decrease in cyst volume. The symptoms caused by increased intracranial pressure related to hydrocephalus, such as headache, nausea, and vomiting, completely relieved in all patients. In the 16 patients who presented visual impairment, visual improvement occurred in 7 patients, completely relieved in 8 patients, and stabilized in one patient after surgery. Limitations of extraocular movement were resolved after the operations, and complete

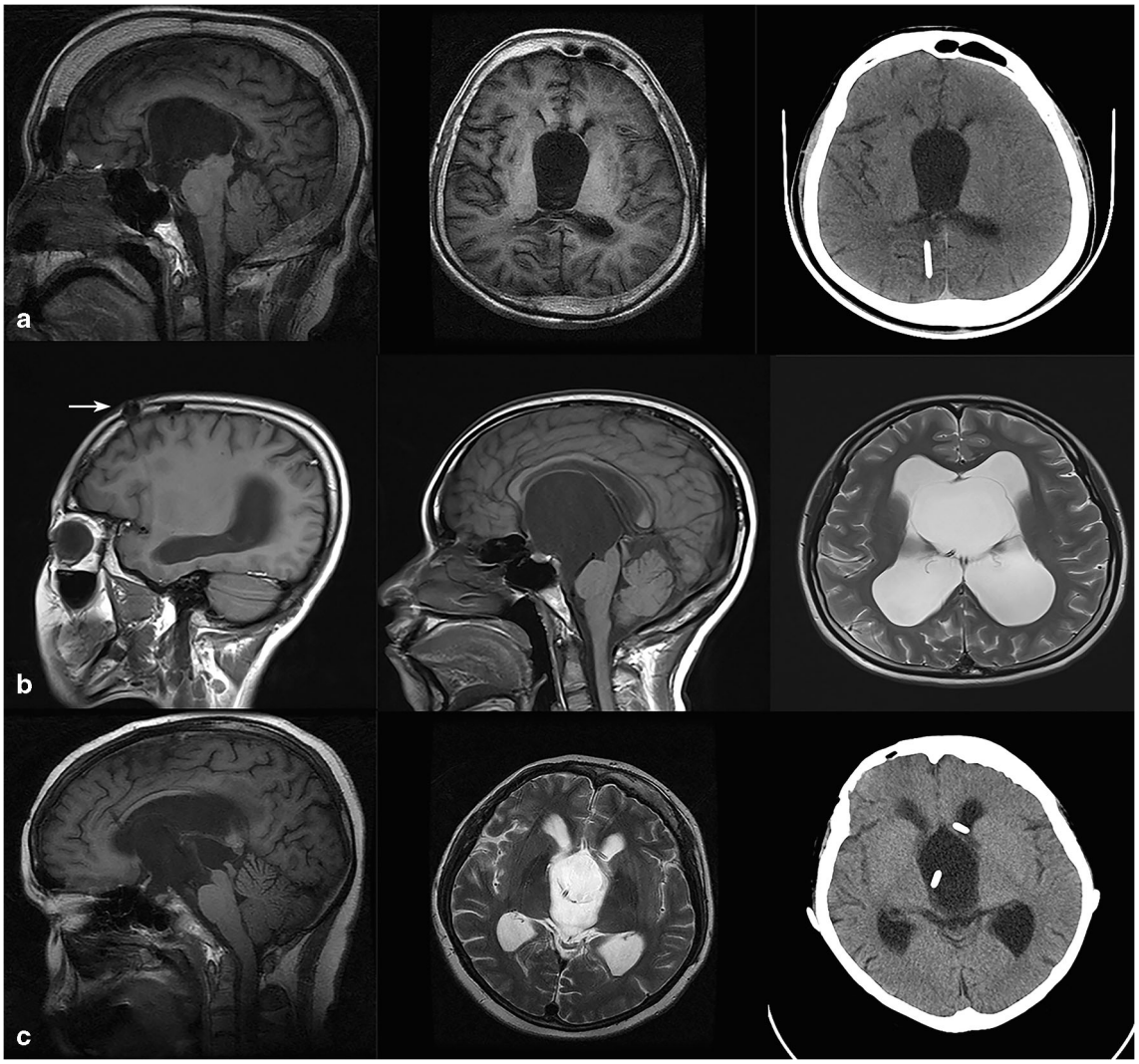

Fig. 1 **a**–**c** Preoperative MRI and CT scans of the patients with a prior VP shunt (cases 9, 13, and 19). Hydrocephalus is not observed in case 9. Arrow indicates the subcutaneous shunt pump

improvement was also observed in the patient with abnormal head movement. In patients with preoperative seizures, they took antiepileptic drug orally for 3 months after surgery and there was no recrudescence after drug discontinuation during the follow-up period. However, the endocrine symptoms and strabismus persisted after operations.

In 23 patients, the cysts decreased in size by 17.8 to 91.2% of their initial volume (mean 63.5 ± 21.2%). The decrease in EI and FOHR ranged from 2.3 to 15.8% (mean 7.89 ± 3.7%) and 3.2 to 13.5% (mean 5.8 ± 2.6%), respectively. A bivariate correlation indicated the irrelevance between the postoperative cyst volume and postoperative EI or FOHR (Pearson correlation: $r = 0.37$, $P = 0.11$; $r = 0.43$, $P = 0.054$) (Fig. 4).

Postoperative complications occurred in two patients: one transient episode of sixth nerve palsy and one subdural hygroma without reoperation.

Illustrative cases

Case 7

A 45-year-old female patient presented with a 7-year history of intermittent headache and a 1-year history of amenorrhea. Over the last 2 years, she had also developed progressive blurring of vision and bilateral visual field defect. MRI showed an oval cyst located at intrasellar and suprasellar region with similar intensity to CSF (Fig. 5f–h). A VCC was performed. Intraoperatively, cyst wall presented a transparent appearance and thin texture (Fig. 5a). There was no obvious slit-valve phenomenon (Fig. 5c). During the follow-up, headache was completely relived and visual impairment was partially improved, but her menstrual pattern was still irregular. Postoperative MRI showed decrease in cyst size (Fig. 5i, j).

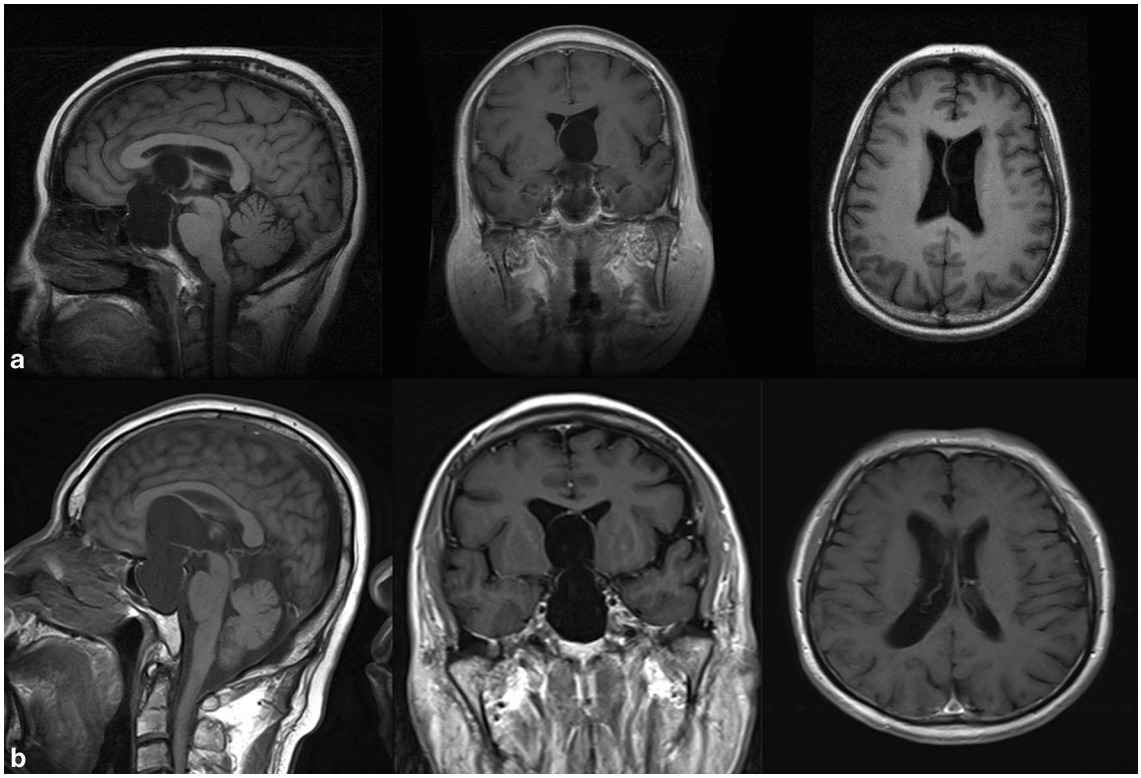

Fig. 2 **a**, **b** Preoperative brain MRI scans of cases 2 and 21 show suprasellar arachnoid cysts without hydrocephalus

Case 19

A 19-year-old female patient visited the outpatient clinic for a 2-month history of headache and visual blurred. When she was 3 years old, she underwent VP shunt because of SAC. Over the last 3 years, she had amenorrhea. Her brain MRI showed that an oval cyst located in suprasellar and interpeduncular cistern with hydrocephalus and a shunt catheter located in right ventricle (Fig. 6e–g). A VCC was performed. Intraoperatively, cyst wall presented a pale appearance and tenacious texture (Fig. 6a). There was no obvious slit-valve phenomenon (Fig. 6c). In addition, the shunt apparatus was removed 3 months after endoscopic fenestration (Fig. 6h). The headache and visual impairment were completely relived, but endocrine disorder persisted after surgery.

Discussions

SACs tend to occur in young children. These lesions account for 11.3% of all intracranial cysts in patients younger than 16 years of age according to the European cooperative study of arachnoid cyst [11]. In our previous report including 73 patients with SACs, 78.1% of patients' age at diagnosis is less than 10 years [8]. In other previous reports, adult patients also always comingle with child patients and only 1–2 adult patients are included. Sood et al. reported 8 cases consisting of 7 children and 1 adult [15]. Gangemi et al. reported 18 arachnoid cysts which included 2 adult SACs [16]. Rappaport reported a 36-year-old patient complained of excessive sleepiness [17]. In addition, a tendency exists to more patients diagnosed in the prenatal period since ultrasonography was used as a prenatal testing tool [3, 18]. These reports well described the clinical presentations, radiological features, treatment modalities, and outcomes of child patients.

SACs could cause clinical symptoms by mass effect and/or obstructing cerebral-spinal fluid (CSF) pathway [19]. Macrocrania and psychomotor retardation were the common symptoms and signs in infants [20]. Symptoms and signs of the intracranial hypertension, psychomotor retardation, short stature, and precocious puberty were more frequent in preschool and adolescent patients [3, 8, 20, 21]. In our adult series, however, the most common clinical presentations were the symptoms related to increased intracranial pressure (61.3%) and visual compromise (54.6%). The optic nerves and chiasm could be stretched by cyst to result into various visual impairments such as unilateral or bilateral decrease in visual acuity or hemianopsia. SACs are a well-known cause of endocrine disorders. Some authors have reported endocrinopathy rates in child patients ranged from 8.9 to 60% [8, 10, 15, 22, 23], but the information on endocrinopathy rates in adult patients were void. Our result revealed the endocrine disorders occurred in 30.4%% ($n = 7$)

Table 1 Summary of clinical data for all patients

Case no.	Age (years)/ sex	Clinical presentations	Endocrine disorder	History of treatment	Hydrocephalus	Endoscopic fenestration	Postoperative visual evaluation	Follow-up (months)
1	44/M	Headache/nausea/vomiting; visual impairment; lower extremity weakness	No	No	Yes	VCC	Partial improvement	28
2	32/M	Headache; visual field defect; memory loss	No	No	No	VC	Complete relief	54
3	19/F	Seizure; instability gait; headache/nausea	No	No	Yes	VCC	–	74
4	49/F	EOM limitation; headache	No	No	Yes	VCC	–	48
5	53/F	Headache/vomiting; visual impairment; lower extremity weakness	No	No	Yes	VC	Complete relief	17
6	43/F	Seizure; instability gait; visual impairment	No	No	Yes	VCC	Partial improvement	48
7	45/F	Headache; visual impairment; hypogonadism	Yes	No	Yes	VCC	Partial improvement	24
8	40/M	Headache; lower extremity weakness	No	No	Yes	VCC	–	60
9	20/F	Microplasia; visual impairment	Yes	VP	No	VCC	Complete relief	54
10	28/F	Headache; visual impairment	No	No	Yes	VCC	Partial improvement	85
11	20/M	Headache; visual impairment; hypogonadism	Yes	No	Yes	VCC	Complete relief	62
12	38/M	Head bobbing	No	No	Yes	VCC	–	36
13	22/M	Headache/vomiting; visual impairment	No	2 VP + open resection	Yes	VCC	Complete relief	42
14	25/F	Headache; visual impairment; hypogonadism	Yes	No	Yes	VCC	Partial improvement	60
15	42/F	Seizure; lower extremity weakness	No	No	Yes	VCC	–	36
16	18/F	Headache; visual impairment	No	No	Yes	VCC	Complete relief	72
17	57/F	Headache/nausea; visual impairment	No	No	Yes	VCC	Stabilization	27
18	26/M	Hypogonadism; visual impairment	Yes	No	Yes	VCC	Partial improvement	81
19	19/F	Headache; visual impairment; hypogonadism	Yes	VP	Yes	VCC	Complete relief	54
20	53/M	EOM limitation; headache	No	No	Yes	VCC	–	48
21	64/M	Headache; visual impairment	No	No	No	VC	Partial improvement	48
22	26/F	Headache; visual impairment; hypogonadism	Yes	No	Yes	VCC	Complete relief	64
23	19/M	Seizure	No	No	Yes	VCC	–	76

F female, *M* male, *EOM* extraocular movement, *VP* ventriculoperitoneal shunt, *VC* ventriculocystostomy, *VCC* ventriculocystocisternostomy

of adult patients. These disorders may be all due to the compressing of the cyst to the hypothalamic–pituitary area and axonal injury caused by hydrocephalus and focal ischemia [24, 25].

The rarity of adult SACs has also prevented an evaluation of characteristics of the cysts and radiological findings. Caemaert et al. were the first to report an endoscopic observation of a slit valve in a SAC [26]. Erşahin et al. reported the slit-valve phenomenon existed in 82.35% of child patients [27]. Fitzpatrick et al. observed the slit-valve mechanism in 83.3% of child patients [28]. In our series, a slit-valve phenomenon was observed in 13 (56.3%) of 23 adult patients, which was significantly lower than in child patients. The result revealed that there could be different mechanisms

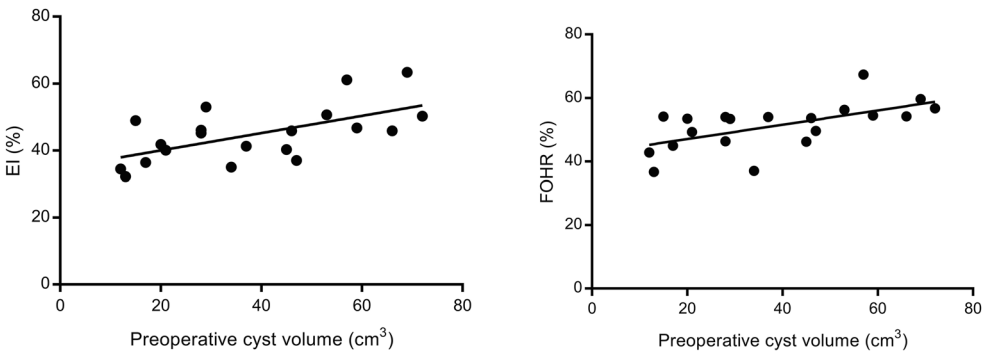

Fig. 3 Scatterplots depicts the correlations between preoperative cyst volume and preoperative EI at presentation (left), and between preoperative cyst volume and preoperative FOHR at presentation (right)

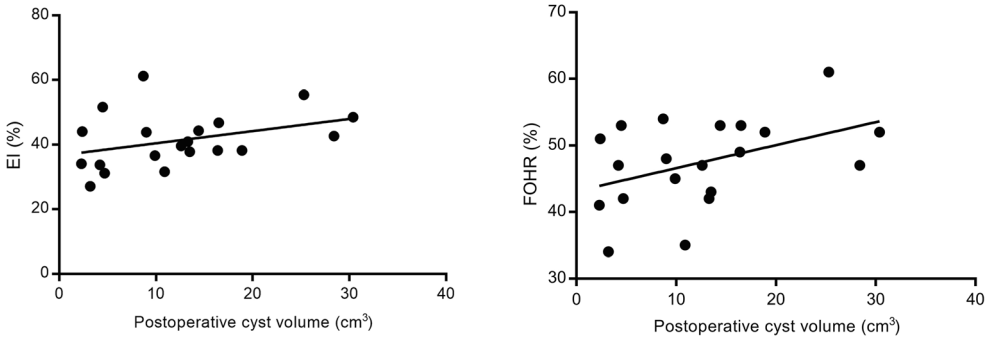

Fig. 4 Scatterplots depicts the correlations between postoperative cyst volume and postoperative EI (left), and between postoperative cyst volume and postoperative FOHR (right)

of cystic dilation between child and adult patients. Crimmins et al. reported the cysts in 21 children ranged in size from 22.4 to 175.9 cm^3 (median 37.1, IQR 27.5–44) at diagnosis [3]. Rizk et al. reported the average cyst size was from 42.98 to 369.2 cm^3 (median 153.96) in 6 child patients [21]. Wang et al. reported the cysts in 6 children ranged in size from 164.8 to 617.6 cm^3 (median 220.6, IQR 187.5–403.9 cm^3) [22]. In our patients, the cyst volumes ranged from 12.3 to 72.5 cm^3 (mean 39.8). These data revealed that a smaller cyst could be frequently observed in adult patients at diagnosis. The possible explanations included limited intracranial compensatory space in adults, different mechanisms of expansion

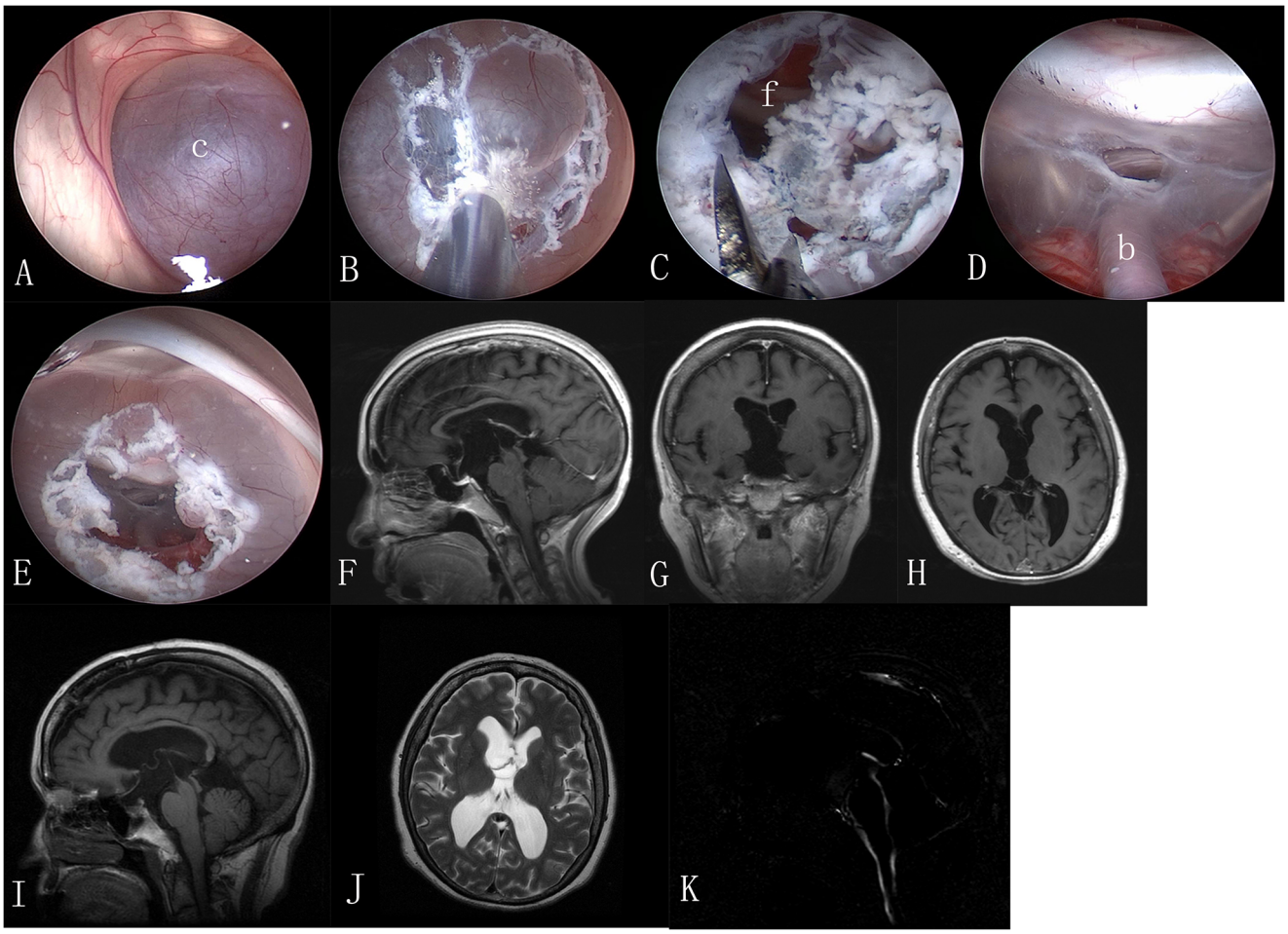

Fig. 5 Case 7. Intraoperative neuroendoscopic views. **a** Cyst protrudes into ventricle through foramen of Monro. Cyst wall presents a transparent appearance and thin texture. **b**, **c** Fenestration of the apical cyst membrane. **d** Fenestration of the basal cyst membrane is made in avascular portions of the basal cyst membrane. **e** After VCC, a close and direct endoscopic view. **f**–**h** Preoperative MRI shows an oval cyst located in suprasellar and interpeduncular cistern with hydrocephalus and a significant compressing of the floor of third ventricle. **i**–**j** Postoperative MR images shows an apparent decrease in the size of the cyst. **k** Cine MRI shows the communication between cistern and cyst. c cyst, f fenestration, b basilar artery

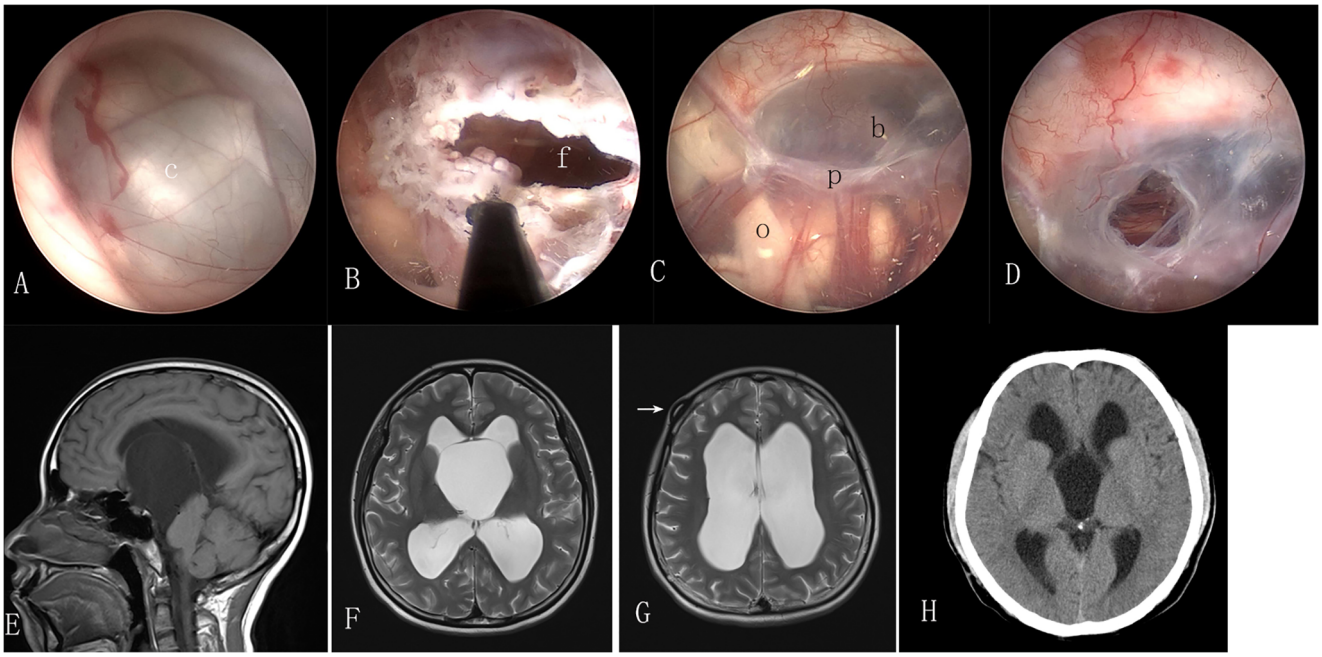

Fig. 6 Case 19. Intraoperative neuroendoscopic views. **a** Cyst protrudes into ventricle through foramen of Monro. Cyst wall presents a pale appearance and tenacious texture. **b** Fenestration of the apical cyst membrane. **c** Anatomical landmarks seen after the endoscope is advanced into the cyst. **d** Fenestration of the basal cyst membrane is made in avascular portions of the basal cyst membrane. **e**–**g** Preoperative MRI shows an oval cyst located in suprasellar and interpeduncular cistern with hydrocephalus and a significant compressing of brain stem and the floor of third ventricle. A prior shunt apparatus locates in right ventricle. Arrow indicates the subcutaneous shunt pump. **h** CT scan 3 years after endoscopic fenestration shows the decrease in cyst size and the shunt apparatus removed. c cyst wall, f fenestration, o oculomotor nerve, b basilar artery, p posterior cerebral artery

and the cumulative effect of synchronous progression of hydrocephalus with cysts, which leaded to adult SACs at diagnosis with a smaller size. Wang et al. reported a mean cyst volume decrease of 52.7% in child patients after fenestration [22]. In the report by Rizk, the average cyst size was 39.92 cm^3 (3.20–101.47) postoperatively [21]. In the report by Crimmins et al., a median decrease in cyst size was 64.7% after VCC [3]. In our patients, a mean decrease in cyst size was 63.5%, which was covered by previous results. We quantitatively evaluated the improvement in hydrocephalus for the first time. The mean decreases in EI and FOHR were 7.89% and 5.8%, respectively. The extent of EI decrease was higher than that of FOHR decrease. The reasonable explanation was that the frontal horn was more susceptible in the condition of obstructive hydrocephalus than occipital horn [29, 30].

Endoscopic fenestration is recommended as the optimal option for SACs due to excellent safety and effectiveness [18, 27, 31–34]. In our series, all patients underwent endoscopic fenestration. During the early period, we choose VC, which is simpler and safer, but as we get more experience, VCC is recommended because it is theoretically considered to be the more effective. In 2011, CormaC et al. performed a meta-analysis of endoscopic fenestration for SACs. Their result revealed the reoperation rate following VC (11 [20%] of 55) was significantly higher than following VCC (9 [9%] of 103; $p = 0.04$) [3]. Until now, there is no single case series has included enough patients to allow for a cogent comparison of patient outcomes following VCC and VC fenestrations. There was no patients who needed reoperation in our series. Slit-valve phenomenon and transparent/thin cyst walls did not influence the treatment outcomes. The endoscopic fenestration is effective to adult SACs.

Limitations

First, because of the retrospective nature of the study, we did not perform further analysis of the difference between pale/tenacious and transparent/thin cyst walls, and we plan to use electron microscope to analyze the difference in the future study. Second, the small sample size limited the power of the results. A study based on a larger cohort is necessary.

Conclusion

Adult SACs are rare. Their typical clinical presentations are not same to the child patients. The most common clinical presentations are the symptoms related to increased intracranial pressure and visual compromise including impairment of visual acuity and visual field, following by endocrine

malfunction. The proportion of slit-valve phenomenon in adult SACs is significantly lower than in child patients, which reveals that there could be different mechanisms of cystic dilation between child and adult patients. The mean cyst volumes in adults at diagnosis are usually smaller than in children. The severity of hydrocephalus is correlated with cyst size. The excellent outcomes in adult SACs can be obtained by endoscopic fenestration.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval The present study was approved by the Ethics Committee of Beijing Tiantan Hospital affiliated to Capital Medical University (Beijing, China).

Informed consent All patients were informed of the purpose of this study and signed a written consent form.

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