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Symptomatic cysts of the cavum septi pellucidi, cavum vergae and cavum veli interpositi: A retrospective duocentric study of 10 patients

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ABSTRACT

Objective: Cysts of the Cavum septi pellucidi (CSP), cavum vergae (CV) and cavum veli interpositi (CVI) are anterior midline intracranial findings which are typically incidental – only rarely do we encounter symptomatic cysts of this type. Only a quite small number of these cysts series have been published, controversies regarding optimal management still exist.

Patients and methods: This is a retrospective study of 10 patients treated at 2 clinics between 2002–2018. 9 patients underwent surgery and 1 is under long-term monitoring. Apart from demographic data, the study analyzed symptoms, cyst size and progression over time, ventricle size, complications, and treatment modality.

Results: CSP with CV was found in 8 cases with 1 case each of CSP and CVI. The study comprised 6 men and 4 women, including 4 children. The mean follow-up time was 43.4 months. The average cyst size was 20.4 mm in CSP and 19.8 mm in CV; the CVI was 33 mm. Headache was most commonly reported (70%) followed by behavioral disturbance (30%). Disturbance in memory, psychomotor development, school performance, visual acuity, and vomiting was variously noted in 20%. The prevailing symptom was headache in adults and behavioral and autonomic disturbance in children. Postoperatively, cysts had reduced by an average of 44.3% while the ventricles remained unchanged. Symptoms resolved in all cases with residual problems in patients presenting with memory loss. No complications were noted.

Conclusion: Endoscopic fenestration is the method of choice in the treatment of symptomatic midline cysts. We recommend that any further research focuses on precisely establishing their clinical presentation, particularly neuropsychological symptoms.

1. Introduction

Cavum septi pellucidi (CSP), cavum vergae (CV) and cavum veli interpositi (CVI) are cavities of the interhemispheric cleavage. These are normal features of intrauterine development and typically close postnatally [1,2]. Even if complete closure does not occur, the remnants of these cavities are asymptomatic in most cases. In rare cases these cavities can become expansive, in which case they are referred to as cysts. Cysts are regarded as symptomatic if compression of structures surrounding an expanding cyst leads to neurological symptoms or obstruction of cerebrospinal fluid (CSF) pathways. The consensus is that the CSP and CV can become symptomatic when over 10 mm in size [3], in which case they are referred to as cysts [2–4]. This is likely true also of CVI [4,5]. Symptoms principally include headaches and hydrocephalus, but also disturbance of development, behavior, memory and visual acuity, as well as macrocrania and epilepsy. Several psychiatric disorders, including schizophrenia have also been linked to these types of cysts [1,3,6,7]. In the past, such lesions were treated by open fenestration, puncture or cystoperitoneal shunting but nowadays endoscopy is considered the method of choice [4–6,8,9]. In this retrospective study we evaluate cases drawn from 2 neurosurgical clinics where endoscopy was performed on symptomatic cysts of the septum pellucidum and velum interpositum. Our principal aim is to highlight a rare and often even underestimated group of illnesses which, where indicated, can be safely and successfully treated endoscopically.

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2. Patients and methods

This is a retrospective study comprising patients from 2 neurosurgical centres (Ostrava and Pilsen) who underwent surgery for symptomatic CSP, CV and CVI cysts between 2002–2018. All patients underwent MR investigation at least once both pre- and postoperation. Endoscopic success and any complications were noted, along with characteristics of individual procedures. In addition to demographic data we also evaluated the duration and character of symptoms, as well as the maximal width of symptomatic cysts and their reduction following surgery. Where possible we attempted to determine any increase in cyst size up to the time of the operation. We also evaluated pre-versus postoperative changes in the maximal width of frontal horns (MWFH) and third ventricle width. Patients were followed up at intervals of 3, 6, 12 and 24 months postoperatively and 12–24 months thereafter. All radiological distances were measured in millimetres using an open-source DICOM viewer (Medixant, Poznan, Poland) by first author of the study. All patients had no history of previous shunting or external drainage implantation.

2.1. Patient history

The study group comprises 9 operated patients and 1 symptomatic patient under long-term monitoring. All patients underwent neurological examination and had been referred by a neurologist to a neurosurgeon. Symptoms, demographic data and radiological findings are summarized in Table 1. Patients are sorted by year of operation. Of the 9 patients admitted, MR investigation demonstrated expansive cysts of the CSP and CV in 8, and an expansive cyst of CVI in 1 patient. Endoscopy was indicated for all patients and all but one underwent surgery. This patient opted for the continuation of a conservative approach despite being symptomatic and was followed up as an outpatient.

2.2. Surgical technique

Endoscopic cyst fenestration involves by definition the creation of a suitably wide connection with the adjacent lateral ventricle (Video 1). An anterior frontal approach was preferred in both clinics this being – among other reasons – the protocol for routinely performing endoscopic third ventriculostomy (ETV). The procedure is similar to ETV, although the entry point is placed more laterally for a directly perpendicular approach to the curved cyst wall. For cysts of the CVI the entry point was shifted laterally and further to the front. We always use neuronavigation for the precise planning of the optimal endoscopic trajectory and all patients were operated on with 0^0 3- or 6-mm endoscopes (Paediscope, Aesculap – B. Braun, Tuttlingen, and Aesculap – B. Braun, Tuttlingen respectively). A right-sided approach was always preferred with respect to individual anatomic variation (proliferation of vascular structures of the septum pellucidum and ventricle walls, or asymmetric curvature of the cyst wall, for example) or the dominant brain hemisphere. Following introduction of the endoscope and revision of lateral ventricle morphology we perform perforation of the septum pellucidum in an avascular area by coagulation. The opening is then dilated by way of forceps or scissors and a Fogarty balloon catheter, which is also employed to control any bleeding from the stoma. If a contralateral ventricle connection is decided upon, this is created through the cyst itself. After confirming hemostasis the endoscope is extracted and sealant patches are applied (TachoSil, Baxter). The burr hole is filled in with the bone dust generated during the craniotomy to maximize the aesthetic result. The procedure is accompanied by the prophylactic intravenous administration of antibiotics (a second or third-generation cephalosporin).

3. Results

Of the 9 cases of CSP, CV was found concurrently in 8. A symptomatic cyst of CVI was determined in 1 patient (Figs. 1 and 2). The study comprised 6 males and 4 females with an average age of 26.2 years (median 22.7 years) ranging from 2 to 69 years. There were 4 pediatric patients making up 40% of the study. CSP cysts averaged 20.4 mm in diameter (median 20 mm), ranging from 13 to 30 mm. CV cysts averaged 19.8 mm (median 16 mm) in diameter ranging from 13 to 29 mm. The CVI cyst was 33 mm. Patients were followed up for an average of 43.4 months (median 46 months), ranging from 3 to 85 months. The average duration of symptoms prior to undergoing the operation was 22.9 months (median 13 months) with the onset of symptoms ranging from 4 to 72 months previously. A right-sided anterior frontal approach was preferred in all cases with 1 exception where a left-sided approach was employed due to the risk of septal vein injury affecting the frontal horn of the right lateral ventricle. Unilateral cyst fenestration was performed in 5 cases (55.6%) and bilateral fenestration in 4 cases (44.4%).

The most common clinical complaint was headaches in 70% of patients, followed by adverse behavioral changes (emotional lability) in 30%. Vomiting, syncope, memory loss, visual disturbance and declining school performance was noted in 20% of study patients. Retarded psychomotor development was observed in 2 pediatric patients. This was particularly apparent in a 2-year-old when compared with his identical twin brother. The other case observed epilepsy, sensorimotor disturbance (including quadriparesis) and progressive macrocephaly. The overriding symptom was headaches, being reported by all adult patients. Other symptoms were represented in one case while behavioral and autonomic disturbances were found respectively in 33.3% of adult patients. In the 4 child patients, behavioral disturbance, delayed psychomotor development and declining school performance were the principal symptoms, being found together in 50% of these cases, while behavioral or autonomic disturbance were noted in 75%. Only 1 child complained of headaches.

Regarding the nature of the headaches, 71.4% (5/7 patients) described acute attacks occurring suddenly or even explosively lasting several minutes and just as quickly subsiding. Between attacks these patients were otherwise asymptomatic. Patient 6 reported chronic and constant dull headaches of fluctuating intensity. ***Patient 8*** was decompensated with the chronic headache with which she had suffered when sudden attacks of severe headache decompensation occurred.

On evaluation of the outcome we found that all patients experienced regression of clinical problems. Headaches, the most frequently occurring complaint, had completely resolved in all operated patients by the time of the initial follow-up at 2–4 months postoperation. No relapse was noted in any of these patients at subsequent follow-ups. Despite the persistence of severe headaches in patient 10 who has been undergoing conservative treatment, this patient is not inclined towards a surgical solution. In the last 2 years the frequency of severe attacks has remained stable at 2–3 times a week.

Symptoms of vomiting, visual disturbance and syncope similarly resolved in the first 3 months postoperatively. In ***patient 4*** with macrocephaly, no further progression in head circumference was observed following the operation. Anterior fontanelle closure occurred in the first year post operation. The delayed psychomotor development observed pre-operatively has since completely resolved. The patient is now developmentally comparable to his identical twin and his school performance has been judged to be above average. Regarding behavioral disturbance or emotional instability, an improvement was noted in adult ***patient 2*** in the first 3 months and in 2 pediatrics (***patients 5 and 7***) in the first 6 months post operation. School performance of these latter two had significantly improved by the end of the first year postoperatively. There has been no recurrence of the epileptic fits experienced by ***patient 7*** since the operation. The memory loss suffered by **patients 7 and 8** has significantly resolved, if not completely: despite improvements they report intermittent short-term memory loss at 21 and 6 months respectively post operation.

Reduction of the maximal cyst width was observed postoperatively

Table 1
Summary of symptoms, demographic data and radiological findings of 10 patients treated at 2 clinics between 2002–2018.

Patient number	Gender	Age (years)	Institution / year of operation	Lesion type	Symptoms	Duration of symptoms (months)	Width of CSP / CV (mm)	Size progression over time	Outcome	Follow-up (months)	Postoperative reduction of width (mm) / (%)
1.	F	22.5	Ostrava / 2002	CSP + CV	Explosive headache.	24	18 / 15	Unknown	Complete recovery	66	10 / 44 (CSP) 9 / 40 (CV)
2.	M	46.8	Ostrava / 2011	CSP + CV	Explosive headache, nausea, vomiting, behavioral disturbance.	4	17 / 14	Unknown	Complete recovery	85	11 / 40 (CSP) 12 / 14 (CV)
3.	M	31	Pilsen / 2011	CSP + CV	Explosive headache.	9	20 / 17	Unknown	Complete recovery	70	9 / 55 (CSP) 8 / 53 (CV)
4.	M	2	Ostrava / 2014	CSP + CV	Progressive macrocephaly, unclosed anterior fontanelle, delayed psychomotor development.	6	20 / 27	CSP 4 mm; CV 5 mm in 3 months	To date nonprogressive macrocephaly; anterior fontanelle fused < 1 year; normal psychomotor development	50	8 / 60 (CSP) 16 / 41 (CV)
5.	M	13.5	Ostrava / 2014	CSP + CV	Progressive behavioral deterioration, uncontrollable mood swings, declining school performance.	23	21 / 15	CSP 9 mm; CV 6 mm in 2 years	Resolution of behavioral disturbance < 6 months	31	7 / 67 (CSP) 11 / 27 (CV)
6.	F	69	Pilsen / 2014	CVI	Progressive deterioration of gait, quadriparesis, headache.	8	33 (CVI)	Unknown	Complete recovery	42	20 / 39 (CVI)
7.	F	9	Ostrava / 2017	CSP + CV	Learning difficulties, unable to concentrate, emotional changes, memory loss, epilepsy, vomiting, loss of consciousness,declining school performance.	72	24 / 28	Unknown	Marked improvements in behavior, learning and concentration; seizure-free since operation; occasional residual short-term memory loss	21	13 / 46 (CSP) 24 / 14 (CV)
8.	F	22.8	Ostrava / 2018	CSP	Headache, vertigo, visual disturbance, memory loss.	17	13	6 months nonprogressive	Complete remission of headache, vertigo and visual disturbance; partial resolution of memory loss	6	7 / 54 (CSP)
9.	M	12.1	Ostrava / 2018	CSP + CV	Explosive headache accompanying collapse, visual disturbance.	6	30 / 13	Unknown	Complete recovery	3	12 / 60 (CSP) 6 / 54 (CV)
10.	M	33	Ostrava / outpatient monitoring	CSP + CV	Progressive headache.	60	21 / 29	CSP 5 mm and CV 8 mm in 5 years	Not operated on	60	Not operated on

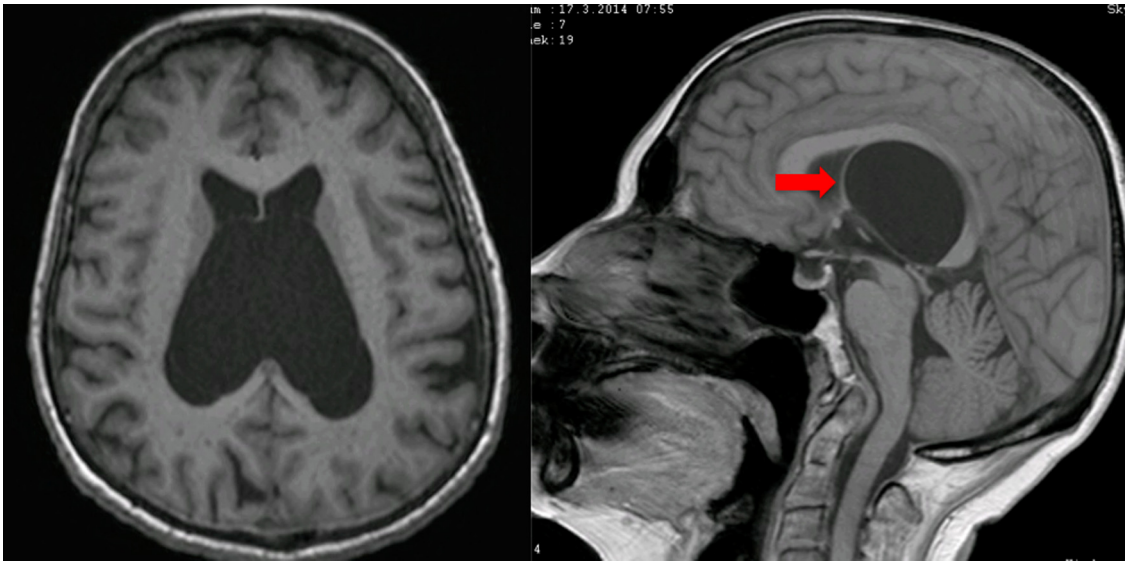

Fig. 1. Patient with cyst of CVI, fornices are elevated anteriorly (red arrow).

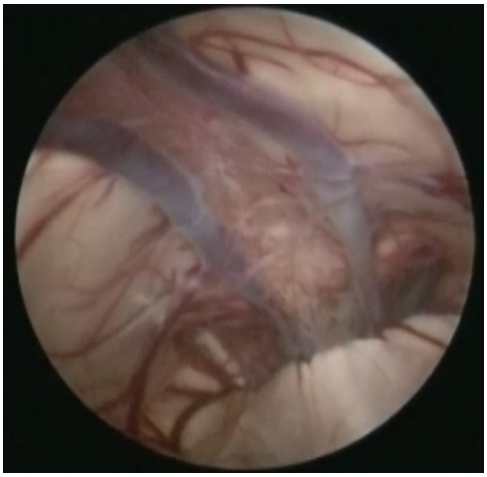

Fig. 2. Internal cerebral veins inside CVI cyst.

in all 9 patients. The CSP of 8 patients was reduced by an average of 53.3% (median 54.5%, range 40–67%), i.e. 10.8 mm (median 11 mm, range 6–18 mm). The CV of 7 patients was reduced by an average of 34.7% (median 40%, range 14–54%), or 6.1 mm (median 6 mm, range 2–11 mm). In the one patient with a cyst of CVI, a reduction of 39% (13 mm) was attained. In adult patients the average reduction in the width of cysts of CSP was 48.3% (median 49%) while in children it was 58.3% (median 60%). Reduction in CV width was similar in adults and children, being 35.7% (median 40%) and 34% (median 34%) respectively. The MWFH and third ventricle width had not significantly changed postoperatively in any patient compared to their preoperative state. The MWFH averaged 36 mm both pre- and post-operatively. Third ventricle width averaged 3.4 mm pre- and 3.7 mm post-operatively. No complications arising as a result of the procedure were noted. Repeated MR examination (at intervals of 3 months to 5 years) was carried out on 4 patients in the study before surgery was indicated: cyst progression was noted in 3 of them (see Table 1).

4. Discussion

Cysts of the CSP, CV and CVI are anterior midline intracranial findings which are typically incidental. A recent study of 19,031 patients (averaging 52.6 years old and ranging from 0 to 99 years) found CSP and CV in 0.93% of cases [10]. CSP with CV was determined in 95.5% of these, while CSP and CV were found independently in 2.8% and 1.7% respectively [10]. According to CT studies, but also confirmed by our own experience, it is clear that CSP almost always occurs in combination with CV – independently-occurring CSP or CV is rare (Fig. 3). Obliteration of these structures begins caudorostrally with the CV from approximately 6 months' gestation, followed by CSP closure typically occurring in the first few months post-natally [1]. It therefore follows that if the process of CV obliteration is interrupted, the CSP will also persist. The isolated occurrence of CV is debatable, considered to be a result of developmental defects in the fornix and splenium of the corpus callosum [3]. The gender specific-incidence has not yet been discussed. In our study, male patients represented 60%. Also, in literature we found a higher incidence of anterior midline intracranial cysts in males. A CT study of 19,031 cases found 177 patients with CSP or CV, with males representing 103 cases (58.2%) and females 74 cases (41.8%) [10]. In separate studies by Wang et al. and Lancon et al, male patients represented 80% [9] and 78% [6] of their respective cohorts. Gangemi et al. describe 4 patients with anterior midline intracranial cysts, of which 3 were male (75%) [4,8]. It seems that incidence of these cysts is more common in males.

Symptomatic cysts typically exhibit lateral bowing of both walls which corrects following fenestration (Fig. 4). Imaging of such cysts in a coronal plane produces characteristic findings which have been dubbed by Libý as 'the wolf sign' [11].

Expanding cysts of the SP become symptomatic through 4 pathophysiological mechanisms: firstly, partial obstruction of the intraventricular foramina by an expanding SP cyst leads to the symptoms of intracranial hypertension (headaches, nausea and vomiting) but can also manifest as sudden collapse and loss of consciousness [6,12]. Another mechanism is deep venous system distortion, as observed by Dandy in one of his patients with marked dilation of the superficial cortical veins of the right hemisphere [6]. Aoki et al. was the first to note the connection between CSP and chronic deep venous impairment [13]. Angiography demonstrated displacement and stretching of the internal cerebral and subependymal veins in a patient with progressive left-sided hemiparesis, a hypotrophic right thalamus and calcification in the head of the right caudate nucleus [13]. We think that congestion in both thalami and the internal capsule caused progressive quadriparesis in our patient with a CVI through compression of the internal cerebral veins. In our 2-year-old patient (patient 4), we pre-operatively observed marked dilation of the superficial cortical veins (Fig. 5) which did not correct postoperation despite regression of symptoms. The question is

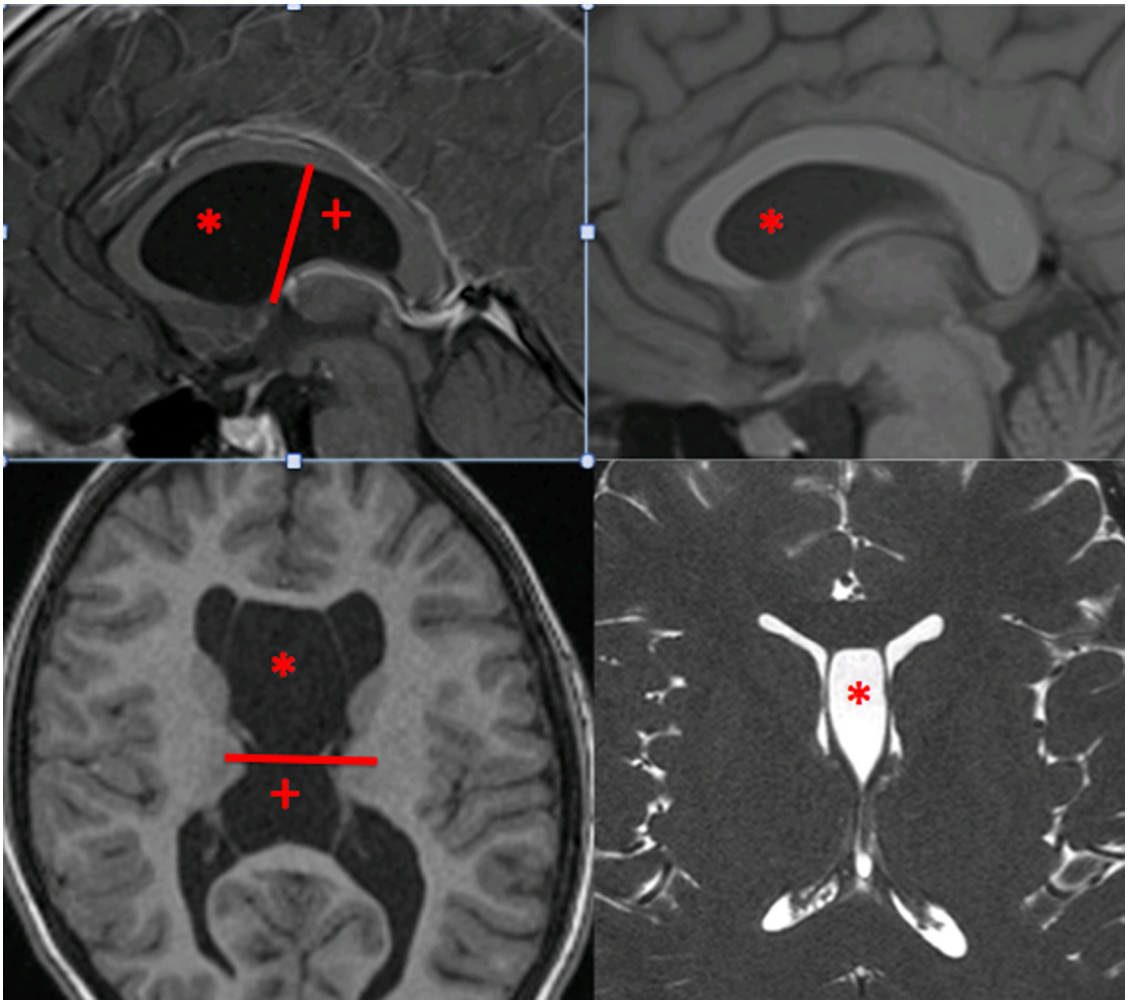

Fig. 3. Images of cysts of CSP (*) and CV (+). Borderline between CSP and CV is marked by red line.

whether this is related to the CSP and CV or if it is an incidental finding. The third possible mechanism is compression of the hypothalamoseptal triangle [6,12]. The symptoms reported in association with this include bizarre behavior, memory loss, developmental delay and learning difficulties in children, disturbed sleep, incontinence and eating disorder [12]. Lastly, compression of visual structures are the result of direct compression by an expansive cyst or hydrocephalus [6,12].

Headaches are most frequent symptom. Pain characteristics described in our study were also commensurate with existing literature, being typically described as acute and sudden, lasting several minutes up to an hour and often induced by physical effort [14]. Lancon et al. evaluated behavioral and autonomic disturbance together which strikes

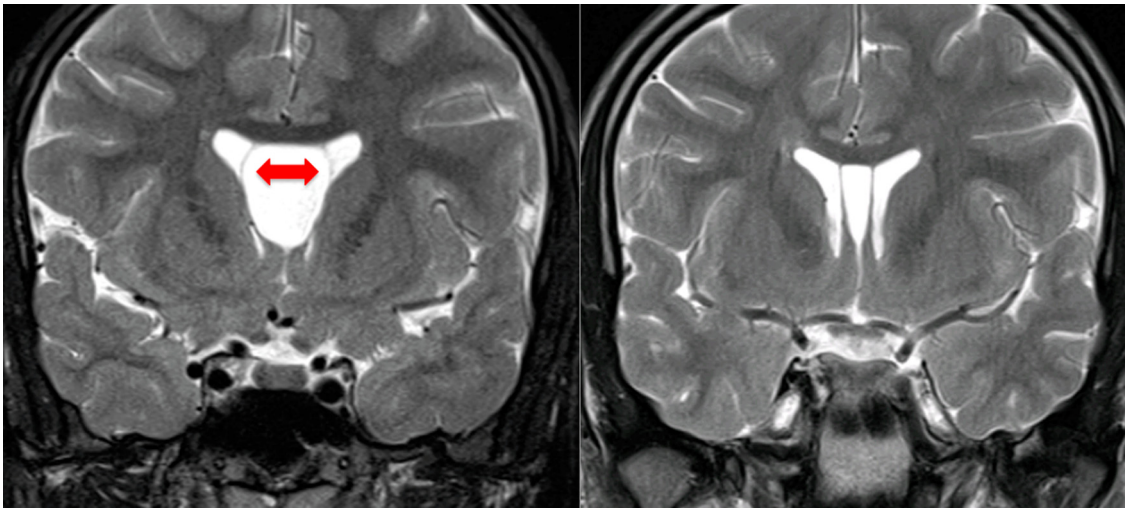

Fig. 4. Symptomatic cysts typically exhibit lateral bowing of both walls (double arrow) which corrects following fenestration.

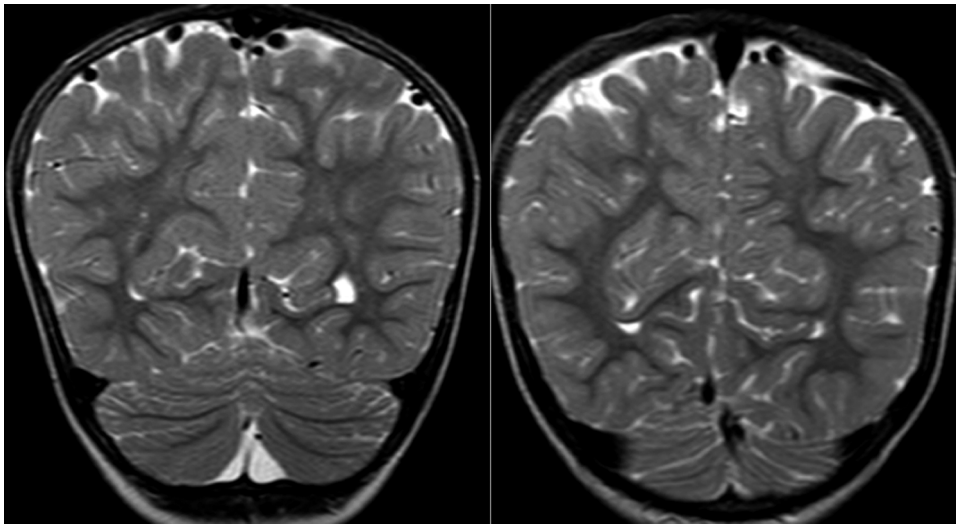

Fig. 5. Patient with marked dilation of the superficial cortical veins. Because of chronic deep venous impairment or only incidental finding?

us as logical given that compression of the hypothalamoseptal triangle is the common underlying cause [6]. Using the same approach we determined such disorders in 50% of the cohort. Interesting results are obtained when analysing incidence of symptoms separately for adults and children. Headache was the dominant symptom among adult patients, being reported in all 6 cases, while behavioral or autonomic disturbance was observed in 33.3%. The pediatric cases most commonly presented behavioral or autonomic disturbance (75%), while headache was reported in just one of them. With Lancon et al, headache was reported in 80% of adult patients and 37.5% of children, while behavioral or autonomic disturbance was observed in 60% of adults and 83% of pediatric cases [6]. Although it would seem that behavioral or autonomic disturbance and headaches tend to be more typical of child and adult patients respectively, we think this is not necessarily the case. For one it is less likely for behavioral changes to become significantly problematic in adult patients. On the other hand, delayed psychomotor development and behavioral changes, especially those affecting educational performance, are more clearly apparent in children. Furthermore, headaches may be difficult to determine in young children, given their inability to accurately express what is bothering them and our reliance on their observations in the evaluation of clinical problems. In such a patient, a headache could present as agitation or other behavioral disturbance wrongly indicating compression of the hypothalamoseptal triangle. However, it is our opinion that these types of cysts are underestimated given their association with neuropsychological disorders. It has been found that patients with schizophrenia and an enlarged CSP (defined as ≥ 7 mm in this study) have lower full scale, verbal and nonverbal IQ scores [15]. Another study demonstrated that patients with large CSP performed worse on verbal comprehension and working memory tests than those with smaller CSP [16]. Thus the question arises whether fenestration should be considered for patients with mental disorder and concurrent large cysts of the CSP, CV or CVI. There are, however, insufficient data at present to make such conclusions.

The majority of CSP, CV, CVI are assumed to freely communicate with the ventricular system (or subarachnoid space in the case of CVI), while symptomatic, expansive cysts are noncommunicating [2,17]. Yamada et al. have demonstrated that an asymptomatic communicative cyst may result in a noncommunicating, expansive and symptomatic cyst [18]. In this particular case, SA bleeding and a communicating CSP (initial CT demonstrated reflux of bleeding into the CSP) developed, 6 months after bleeding, into severe headaches alongside progressive widening of the CSP and ventricles [18]. Yamada et al. present a new MR technique – time-spatial labelling inversion pulse (time-SLIP) – for the demonstration of a noncommunicating CSP prior to endoscopic fenestration [18]. This imaging modality can also be utilized post-operation to confirm successful endoscopic fenestration by, conversely, demonstrating communication between the CSP and ventricles [18]. Although we have no experience with this imaging modality, it appears to be very advantageous in the conclusive diagnosis of a non-communicating CSP.

History of the treatment of these lesions is summarized in Table 2 [4–6,8,13,19–32]. Jackowski et al. were the first to describe successful cyst fenestration by endoscopy in 1995 [23]. Nowadays, this is considered a safe and routine method in the treatment of cysts of CSP, CV and CVI [4,5,8,9,12], with improvement of symptoms reported in all the above-mentioned studies. In neuroendoscopic procedures, the reported mortality and the morbidity is below 2% respectively 1% [33,34]. Minor fornix contusion is the most common, but usually asymptomatic intraoperative complication without any memory consequences [34].

In the case of cysts of CSP and CV the optimal approach remains controversial: while the majority of authors prefer a frontal approach [9,19,35], Gangemi et al. recommend a posterior approach from the right occipital horn to the cyst [8]. The Italian authors suggest this approach as it enables the easier cannulation of the occipital horn, which is usually larger than the frontal one thus avoiding the risk of damage to vascular and neural structures [8]. Furthermore, it provides a more direct endoscopic trajectory [8]. With cysts of the CVI, both frontal and posterior approaches have been employed [4,5,12]. In our study a right frontal approach was always preferred with an exception made in 1 case, where a left-sided approach was employed due to the risk of septal vein damage affecting the right frontal horn. With the assistance of neuronavigation we did not encounter problems penetrating the frontal horn, even though it is true that the occipital horn is, in most cases, wider and its targeting potentially easier. In our opinion the use of neuronavigation is essential, not the type of approach. For identification of safe trajectory of neuroendoscopic procedures around eloquent areas is recommended the use of preoperative navigated transcranial magnetic stimulation and tractography [36].

It is also disputed whether unilateral or bilateral fenestration (i.e. fenestration through the cyst into the contralateral ventricle) should be performed [8,9,19,35]. The advantage of the bilateral approach is that it improves the chances of long-term patency [8]. Despite this assertion, we have no personal experience of relapse in cases of such cysts, nor could we find any mention of this in the literature. On the other hand it is sensible not to completely rule out the possibility, as for example, in ETV. Wang et al, in a study of 14 successfully-treated patients,

Table 2
Outcome and treatment in series of symptomatic anterior midline intracranial cysts.

Author, Year	Age (years) / Sex	Type of cyst	Treatment	Outcome, follow up
Dandy, 1931	50 / M	CSP/CV	craniotomy	died
	4 / M	CSP/CV	craniotomy	improved, 8 months
Miller, 1949	35 / M	CSP	percutaneous puncture	improved, 1 year
Aboulker, 1968	40/ F	?	craniotomy	improved, 10 months
Wilson & Howieson, 1970	61 / F	CSP	craniotomy	improved, 1 year
Heiskanen, 1973	22 months / F	CSP	craniotomy	improved, 1 year
Gubbay, 1977	42 / M	CSP	craniotomy	recurrence in one year, after repeated craniotomy asymptomatic, 22 years
	61/ M	CSP	craniotomy, cysto-ventriculo-atrial shunt	recurrence in 8 months, after shunt asymptomatic, 15 years
Cowley, 1979	52 / M	CSP / CV	craniotomy	improved, 10 months
Amin, 1986	14 / M	CSP	craniotomy	improved, 17 months
Aoki, 1986	11/ M	CSP	craniotomy	stable, 1 year
Donauer, 1986	11/ F	CV	stereotactic internal shunt (cysto-ventricular)	improved, follow up not specified
	36 / F	CV	stereotactic internal shunt (cysto-ventricular)	improved, 3 years
	63 / M	CV	stereotactic internal shunt (cysto-ventricular)	improved, 2 years
	22 / F	CV	stereotactic internal shunt (cysto-ventricular)	improved, 6 months
Wester, 1990	2,5 / M	CSP/ CV	stereotactic internal shunt (cysto-ventricular)	improved, 1 year
	8 / M	CSP / CV	stereotactic internal shunt (cysto-ventricular)	improved, follow up not specified
Silbert, 1993	60 / F	CSP	stereotactic puncture	improved, 3 years
	6 months / M	CSP	ventriculo-peritoneal shunt	died
Miyamory, 1995	6 / M	CSP / CV	stereotactic ventriculo-peritoneal shunt	improved, follow up not specified
Jackowski, 1995	60 / M	CSP	endoscopic	improved, 5 months
Lancon, 1996	8 / M	CSP	cysto-peritoneal shunt	improved, 1 year
Gangemi, 1997	9 / M	CVI	endoscopic	improved, follow up not specified
Gangemi, 2002	8 / M	CSP	endoscopic	improved, 15 months
	7 / M	CSP	endoscopic	improved, 6 months
	21 / M	CSP	endoscopic	improved, 1 month
Fratzoglou, 2003	48 / F	CSP / CV	endoscopic	improved, 4 years
	64 / F	CSP / CV	endoscopic	improved, 6 months
	34 / M	CSP / CV	endoscopic	improved, 3 months
	44 / M	CSP / CV	endoscopic	improved, 3 years
Tong, 2012	3 / M	CVI	endoscopic	improved, 6 months
	13 / F	CVI	endoscopic	partially improved, 2 years
Wang, 2013 (14 patients)	8 – 38 / 11M & 3F	CSP	endoscopic	all improved, follow up 6 months – 2 years

confirmed that unilateral fenestration is sufficient [9]. We agree that any attempt at bilateral fenestration should be performed bearing in mind the higher risk of damaging neural and vascular structures. The septal veins, for example, may not be easily visualized from the interior wall of the cyst. In our study, bilateral fenestration was performed in 5 cases and unilateral fenestration in 4; no complications were encountered in connection with either technique. We are of the opinion that unilateral fenestration is a fully satisfactory technique.

All successfully treated cysts in literature underwent reductions in their volume and size, although exact quantification of these changes is lacking [4,8,9,12]. In our study, CSP cyst reduction was, on average, higher among children, while CV cyst reduction was broadly similar between adults and children. We were also interested in establishing if there was any postoperative reduction in the ventricular system: no change in ventricular width was noted among our patients. This results are similar to those of Wester et al, who observed postoperative CSP and CV cyst reduction while ventricular width remained unchanged [17].

5. Conclusion

Endoscopic fenestration is the method of choice in the safe and successful treatment of symptomatic midline cysts. We recommend that additional research focuses on the detailed analysis of their clinical presentation, particularly regarding neuropsychological symptoms – these have not been precisely established and can be overlooked.

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Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (name of institute/committee) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in the study

Declaration of Competing Interest

None.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.clineuro.2019.105494.

References

[1] A. Yokota, Cysts originating from a defect in the hemispheric cleavage (Cavum septi pellucidi, cavum vergae, cavum veli interpositi), in: A.J. Raimondi, M. Choux, C. Di Rocco (Eds.), Intracranial Cyst Lesions, Springer, New York, New York, NY, 1993, pp. 87–99, , https://doi.org/10.1007/978-1-4615-7281-7_6.

[2] C.M. Shaw, E.C. Alvord, Cava septi pellucidi et vergae: their normal and pathogical states, Brain J. Neurol. 92 (1969) 213–223.

[3] M. Sarwar, The septum pellucidum: normal and abnormal, AJNR Am. J. Neuroradiol. 10 (1989) 989–1005.

[4] M. Gangemi, P. Donati, F. Maiuri, L. Sigona, Cyst of the velum interpositum treated by endoscopic fenestration, Surg. Neurol. 47 (1997) 134–136 discussion 136–137.

[5] C.K.W. Tong, A. Singhal, D.D. Cochrane, Endoscopic fenestration of cavum velum interpositum cysts: a case study of two symptomatic patients, Childs Nerv. Syst. ChNS 28 (2012) 1261–1264, https://doi.org/10.1007/s00381-012-1770-4.

[6] J.A. Lancon, D.E. Haines, F.A. Raila, A.D. Parent, V.V. Vedanarayanan, Expanding cyst of the septum pellucidum. Case report, J. Neurosurg. 85 (1996) 1127–1134, https://doi.org/10.3171/jns.1996.85.6.1127.

[7] C. Trzesniak, I.R. Oliveira, M.J. Kempton, A. Galvão-de Almeida, M.H.N. Chagas, M.C.F. Ferrari, A.S. Filho, A.W. Zuardi, D.A. Prado, G.F. Busatto, P.K. McGuire, J.E.C. Hallak, J.A.S. Crippa, Are Cavum Septum Pellucidum Abnormalities More Common in Schizophrenia Spectrum Disorders? A Systematic Review and Meta-Analysis, (2011), https://doi.org/10.1016/j.schres.2010.09.016.

[8] M. Gangemi, F. Maiuri, P. Cappabianca, C. Alafaci, O. De Divitiis, F. Tomasello, E. De Divitiis, Endoscopic fenestration of symptomatic septum pellucidum cysts: three case reports with discussion on the approaches and technique, Minim. Invasive Neurosurg. 45 (2002) 105–108, https://doi.org/10.1055/s-2002-32483.

[9] L. Wang, S.Y. Ling, X.M. Fu, C.S. Niu, R.B. Qian, Neuronavigation-assisted endoscopic unilateral cyst fenestration for treatment of symptomatic septum pellucidum cysts, J. Neurol. Surg. Part Cent. Eur. Neurosurg. 74 (2013) 209–215, https://doi.org/10.1055/s-0032-1331385.

[10] J.-J. Chen, C.-J. Chen, H.-F. Chang, D.-L. Chen, Y.-C. Hsu, T.-P. Chang, Prevalence of cavum septum pellucidum and/or cavum Vergae in brain computed tomographies of Taiwanese, Acta Neurol. Taiwan. 23 (2014) 49–54.

[11] P. Liby, Letter: the wolf sign, Neurosurgery 81 (2017) E68–E69, https://doi.org/10.1093/neuros/nyx460.

[12] P. Donati, L. Sardo, M. Sanzo, Giant cyst of the cavum septi pellucidi, cavum Vergae and veli interpositi, Minim. Invasive Neurosurg. MIN 46 (2003) 177–181, https://doi.org/10.1055/s-2003-40732.

[13] N. Aoki, Cyst of the septum pellucidum presenting as hemiparesis, Childs Nerv. Syst. ChNS 2 (1986) 326–328.

[14] K.C. Wang, J.L. Fuh, J.F. Lirng, W.C. Huang, S.J. Wang, Headache Profiles in Patients With a Dilatated Cyst of the Cavum Septi Pellucidi, (2004), https://doi.org/10.1111/j.1468-2982.2004.00760.x.

[15] P. Nopoulos, A. Krie, N.C. Andreasen, Enlarged cavum septi pellucidi in patients with schizophrenia: clinical and cognitive correlates, J. Neuropsychiatry Clin. Neurosci. 12 (2000) 344–349, https://doi.org/10.1176/jnp.12.3.344.

[16] L.A. Flashman, R.M. Roth, H.S. Pixley, H.B. Cleavinger, T.W. McAllister, R. Vidaver, A.J. Saykin, Cavum septum pellucidum in schizophrenia: clinical and neuropsychological correlates, Psychiatry Res. 154 (2007) 147–155, https://doi.org/10.1016/j.pscychresns.2006.09.001.

[17] K. Wester, J. Kråkenes, G. Moen, Expanding cava septi pellucidi and cava vergae in children: report of three cases, Neurosurgery 37 (1995) 134–137.

[18] S. Yamada, T. Goto, J.G. Mccomb, Use of a Spin-Labeled Cerebrospinal Fluid Magnetic Resonance Imaging Technique to Demonstrate Successful Endoscopic Fenestration of an Enlarging Symptomatic Cavum Septi Pellucidi, (2013), https://doi.org/10.1016/j.wneu.2012.01.039.

[19] M. Fratzoglou, P. Grunert, A. Leite dos Santos, P. Hwang, G. Fries, Symptomatic cysts of the cavum septi pellucidi and cavum vergae: the role of endoscopic neurosurgery in the treatment of four consecutive cases, Minim. Invasive Neurosurg. MIN 46 (2003) 243–249, https://doi.org/10.1055/s-2003-42351.

[20] J. Aboulker, P. Pradat, J. Metzger, L. Harispe, N. Dodan, B. Bringer, Intracranial hypertension due to septal cyst blocking Monro's foramen, Neurochirurgie 14 (1968) 901–907.

[21] T. Miyamori, K. Miyamori, T. Hasegawa, K. Tokuda, Y. Yamamoto, Expanded cavum septi pellucidi and cavum vergae associated with behavioral symptoms relieved by a stereotactic procedure: case report, Surg. Neurol. 44 (1995) 471–475.

[22] K. Wester, P.H. Pedersen, J.L. Larsen, P.E. Waaler, Dynamic aspects of expanding cava septi pellucidi et Vergae, Acta Neurochir. (Wien) 104 (1990) 147–150, https://doi.org/10.1007/BF01842833.

[23] A. Jackowski, M. Kulshresta, S. Sgouros, Laser-assisted flexible endoscopic fenestration of giant cyst of the septum pellucidum, Br. J. Neurosurg. 9 (1995) 527–531.

[24] E. Donauer, J.R. Moringlane, C.B. Ostertag, Cavum vergae cyst as a cause of hydrocephalus, "almost forgotten"? Successful stereotactic treatment, Acta Neurochir. (Wien) 83 (1986) 12–19.

[25] B.H. Amin, Symptomatic cyst of the septum pellucidum, Childs Nerv. Syst. ChNS 2 (1986) 320–322.

[26] D. Miller, Cyst of the fifth ventricle treated by intraventricular drainage, J. Neurosurg. 6 (1949) 332, https://doi.org/10.3171/jns.1949.6.4.0332.

[27] C.B. Wilson, J. Howieson, Cysts of the septum pellucidum. Review and report of one case, Neurochirurgia (Stuttg.) 13 (1970) 93–99, https://doi.org/10.1055/s-0028-1095263.

[28] O. Heiskanen, Cyst of the septum pellucidum causing increased intracranial pressure and hydrocephalus. Case report, J. Neurosurg. 38 (1973) 771–773, https://doi.org/10.3171/jns.1973.38.6.0771.

[29] P.L. Silbert, S.S. Gubbay, R.J. Vaughan, Cavum septum pellucidum and obstructive hydrocephalus, J. Neurol. Neurosurg. Psychiatry 56 (1993) 820–822, https://doi.org/10.1136/jnnp.56.7.820.

[30] A.R. Cowley, D.M. Moody, E. Alexander, M.R. Ball, D.W. Laster, Distinctive CT appearance of cyst of the cavum septi pellucidi, AJR Am. J. Roentgenol. 133 (1979) 548–550, https://doi.org/10.2214/ajr.133.3.548.

[31] W. Dandy, Congenital Cerebral Cysts of the Cavum Septi Pellu- Cidi (Fifth Ventricle) and cavum Vergae (Sixth Ventricle) 25 (1931), pp. 44–66.

[32] S.S. Gubbay, R. Vaughan, J.S. Lekias, Intermittent hydrocephalus due to cysts of the septum pellucidum: a study of three cases, Clin. Exp. Neurol. 14 (1977) 93–99.

[33] H.W.S. Schroeder, J. Oertel, M.R. Gaab, Incidence of complications in neuroendoscopic surgery, Childs Nerv. Syst. ChNS 20 (2004) 878–883, https://doi.org/10.1007/s00381-004-0946-y.

[34] J. Oertel, S. Linsler, C. Emmerich, D. Keiner, M. Gaab, H. Schroeder, S. Senger, Results of combined intraventricular neuroendoscopic procedures in 130 cases with special focus on fornix contusions, World Neurosurg. 108 (2017) 817–825, https://doi.org/10.1016/j.wneu.2017.09.045.

[35] T. Miki, J. Wada, N. Nakajima, T. Inaji, J. Akimoto, J. Haraoka, Operative indications and neuroendoscopic management of symptomatic cysts of the septum pellucidum, Childs Nerv. Syst. ChNS 21 (2005) 372–381, https://doi.org/10.1007/s00381-004-1063-7.

[36] P. Hendrix, S. Senger, C.J. Griessenauer, A. Simgen, S. Linsler, J. Oertel, Preoperative navigated transcranial magnetic stimulation and tractography to guide endoscopic cystoventriculostomy: a technical note and case report, World Neurosurg. 109 (2018) 209–217, https://doi.org/10.1016/j.wneu.2017.09.185.