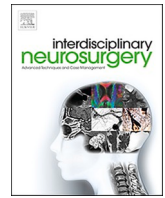




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Research Article

Surgical management of ruptured intracranial arachnoid cysts in young adults: the role of subduroperitoneal shunt placement

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ABSTRACT

Introduction: Intracranial arachnoid cysts (IAC) are a common incidental finding on brain MRIs in adults. While IAC rupture, leading to severe intracranial hypertension, is well-documented in children, it is rarely reported in adults. As a result, there is limited guidance on managing ruptured IACs in young adults.

In particular, there is no data on the benefits of subduroperitoneal (SDP) shunt placement without interposed pressure valves in young adult patients, a treatment often used in pediatric patients.

Methods: We present two cases of young adult patients presenting with ruptured IAC with subsequent subdural hematoma/collection and consecutive neurological decline. Based on these sample clinical cases, we will discuss the above-mentioned management strategy and provide an overview of the current available literature.

Results: This case series demonstrates the successful management of ruptured IAC in young adults through drainage of subdural fluid collections via burr hole trepanation, combined with SDP shunt placement without interconnected pressure valves.

Conclusion: While there is general consensus that surgery is necessary for symptomatic cases following IAC rupture, the optimal approach remains debated. We provide additional evidence that burr hole trepanation combined with SDP shunt placement without interconnected pressure valves, a method commonly used in children, is a safe treatment option for young adults with ruptured IAC.

1. Introduction

Intracranial arachnoid cysts (IAC) account for 1 %–2.5 % of all intracranial masses, with prevalence varying depending on the population studied [8,13,21] to [17,20]. There is a notably higher prevalence of IAC in the pediatric age group compared to adults [22]. Although the pathogenesis of IAC is not well understood, most cases are sporadic, with rare familial forms reported. IAC show a male predominance, and are most commonly located around the Sylvian fissure or the middle cranial fossa with a tendency towards the left [4,8,15,16,18,21].

In adults, IACs are often asymptomatic, and it is estimated that 75 % of symptomatic cases present in childhood [16,19,21]. When symptomatic, IACs may cause a range of symptoms depending on their

location, growth rate, and space-occupying effects [4,7,9,10,13,16].

Rupture of an IAC, leading to subdural hematoma or fluid collection and resulting in severe intracranial hypertension, is a serious complication well-documented in pediatric patients. However, such complications are rarely described in adults. Consequently, there is a lack of literature on the appropriate management of ruptured IAC in young adults. Specifically, there is no current data on the efficacy of subduroperitoneal (SDP) shunt placement without interposed pressure valves in this age group, despite its frequent use in pediatric cases.

2. Materials & methods

We will examine the proposed surgical approach by presenting two

Abbreviations: CSF, Cerebrospinal fluid; CP, Cystoperitoneal; FLAIR, Fluid-attenuated inversion recovery; cMRI, Cranial Magnetic Resonance Imaging; IAC, Intracranial arachnoid cysts; ICP, Intracranial pressure; OCT, Optical coherence tomography; POD, Postoperative day; SAC, Sylvian arachnoid cysts; DP shunt, Subduroperitoneal shunt.

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clinical case studies. Additionally, we will provide an overview of the existing literature on ruptured IAC to contextualize this management strategy.

3. Results

3.1. Historical background

While the triggers and risk factors for the rupture of intracranial arachnoid cysts (IACs) are still debated, the most widely accepted theory posits that a prior mild head trauma may lead to tearing of the bridging veins [16,20]. Rarely, spontaneous events have been described [3,13,14,16,20].

Massimi et al. [16], including their own series of 16 children and an additional 430 young patients from the literature, recently presented a rupture risk calculation specifically for Sylvian arachnoid cysts (SACs), which represent the largest category of IACs [4,7,16]. They reported an overall rupture risk of 0.04 %, with a higher risk of up to 10 % in children with SAC [16].

Currently, the surgical management of IACs lacks consistent evidence [4,8,19,20]. There is general consensus that emergency surgery is required for symptomatic patients with IAC rupture [4,16,18,19]. However, the choice of surgical approach remains controversial [4,8,20,21]. The decision is typically influenced by factors such as the location of the cyst, its compressive effects [1,3,4,11,13], potential disturbances in cerebrospinal fluid dynamics [19], and the surgeon's preference.

3.2. Clinical presentation

The typical clinical presentation of an intracranial arachnoid cyst (IAC) rupture is associated with increased intracranial pressure (ICP) and is radiologically correlated with underlying subdural hematoma or fluid collection [7,16,20]. In young adults, although rare, IAC rupture should be considered as a potential cause of headache [3,4,7,18,20], as well as other non-specific symptoms of intracranial hypertension [3,4,16,17,21] and seizures [17,21]. These symptoms are among the most commonly reported. In contrast, baseline conditions in young adults differ from those in older age groups, where regular medical consultations for underlying conditions are more prevalent and both patients and healthcare professionals are generally more aware of potential issues. For instance, migrainous symptoms, as seen in one of our cases, are relatively common in younger patients and can complicate the diagnostic process by masking the symptoms of IAC rupture. In the elderly, symptoms may resemble those of subdural hematoma or normal pressure hydrocephalus, including dementia, urinary incontinence, or hemiparesis [22].

3.3. Diagnosis, management

While there is a general consensus that surgery is necessary for symptomatic cases following IAC rupture, the optimal surgical target and approach remain contentious, with several surgical strategies proposed. These approaches include direct intervention on the ruptured IAC, either through microsurgical or endoscopic fenestration, and optional marsupialization or complete cyst resection via an open surgical approach [2,3,8,11]. Some protocols also include the placement of a cystoperitoneal (CP) shunt [2,3,7,13,15]. Alternatively, cystocisternostomy may be considered, such as connecting a ruptured Sylvian arachnoid cyst with the basal cisterns or employing a trans-aqueductal *trans-Magendie* approach specifically for IACs in the posterior fossa [11]. On the other hand, the resulting subdural component after cyst rupture can be addressed *per se*. Strategies for fluid drainage include burr hole trepanation alone [12], or sequential drainage followed by SDP shunt placement, particularly in pediatric patients [1,6].

Few studies have specifically investigated the management of adult

patients with ruptured IACs [1,3,5,17,18,22]. These studies primarily compared resection or fenestration techniques, often combined with CP shunts, which address the cyst itself [3,5,17,18,22]. Some studies included mixed pediatric and adult populations [1,3,18], or focused on different locations such as spinal arachnoid cysts [18], limiting their applicability to young adults with ruptured IACs. For example, Fuentes et al. [12] successfully treated three adult patients using burr hole trepanation alone for subdural collection. Albuquerque [1] did not specify the technique for draining underlying subdural fluid collections in their cohort of five patients, which included both pediatric and adult cases, although they reported significant resorption of subdural hematomas and reduction in cyst size in three out of five cases. In the remaining two cases, where the IAC rupture extended into the basal cisterns, neither the cyst nor the subdural collection showed significant size changes.

3.4. Prognosis and outcomes

It is crucial to acknowledge that complications arising from the rupture of an IAC can progress swiftly [16]. In cases where a subdural collection is identified as the primary cause of rapid neurological decline following cyst rupture, we recommend focusing on the management of the subdural collection rather than the cyst itself in the acute setting. The IAC itself has most likely been present since prenatal development [7,17], allowing patients a substantial period to adapt to the space-occupying effect of the cyst, despite its often notable size [17].

Moreover, the risk of postoperative complications following cyst fenestration is relatively high [15]. Postoperative subdural hematoma is the most frequent complication, occurring in 4.9 % of cases, with similar rates for both endoscopic and microsurgical techniques [15]. Additionally, cyst resection or marsupialization necessitates larger surgical approaches through extended skin incisions, which inherently pose an increased risk of blood loss, infection, and necrosis compared to our method. We believe that these approaches could be avoided, as they may expose an otherwise asymptomatic patient to unnecessary risk.

3.5. Exemplary case description

In this case series, we illustrate the successful outcomes achieved through SDP shunting, without the need for additional pressure valves, in two young adults with ruptured arachnoid cysts at our institution.

3.5.1. Case 1

A 29-year-old male patient presented with cephalgia and diplopia, referred to our facility for further evaluation. His medical history was otherwise unremarkable, except for corrected bilateral myopia (−3/−3.5 diopters). The symptoms began three weeks prior and were characterized by band-shaped headaches triggered by rapid movements, predominantly on the right side, radiating retroorbitally and nuchally. The pain was described as stabbing. Neurological examination revealed horizontal diplopia, slight gait ataxia, and upper limb dysmetria, but was otherwise intact. No signs of infectious disease were noted upon internal examination. The patient, who worked full-time as a physicist and had previously lived independently, experienced significant limitations in daily activities due to visual disturbances, resulting in a Karnofsky Performance Score of 60 %.

Cranial magnetic resonance imaging (cMRI) revealed a temporosylvian arachnoid cyst measuring 9 × 7 × 9 cm, with associated bihemispheric fluid formations measuring 9 mm on the left and 5 mm on the right (Fig. 1 A/B). These findings led to a diagnosis of a ruptured left temporal arachnoid cyst with a resultant midline shift to the right and space-occupying bihemispheric hygroma. Neuro-ophthalmological examination demonstrated right-sided abducens nerve palsy and bilateral papilledema. To address elevated intracranial pressure (ICP), an SDP shunt was promptly implanted. The SDP system included a shortened ventricular catheter directed towards the left frontal subdural collection,

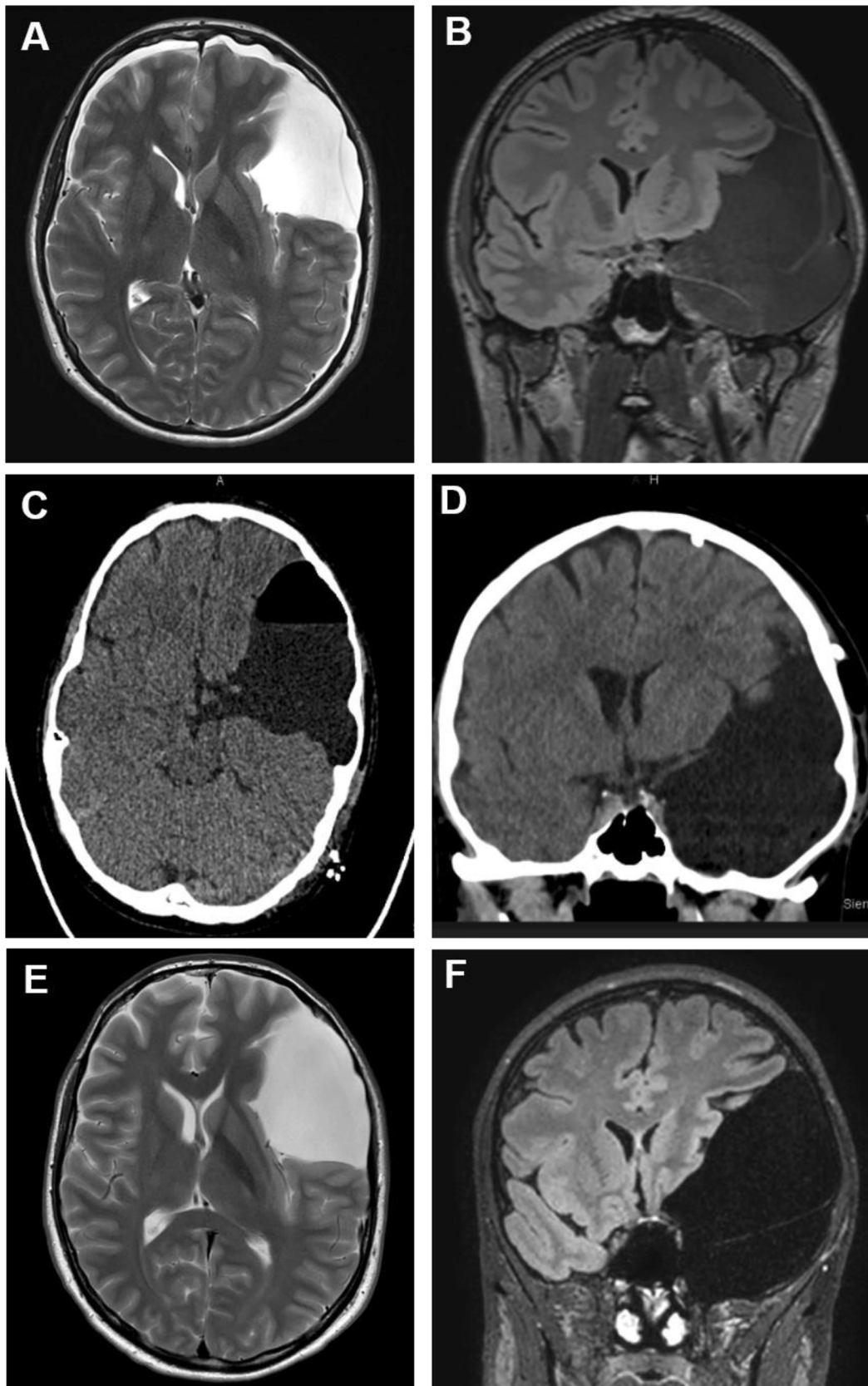


Fig. 1. Axial T2 section reveals the extensive, extraaxial CSF isointense space-occupying lesion measuring 9x7x9cm in the left medial and anterior cranial fossa with bihemispheric fluid collections (A). There is a resulting midline shift of 2–3 mm to the right. Coronal T2 Flair section shows the ruptured arachnoid cyst with detachment of the cyst from the dura (B). Axial (C) and cranial (D) section of CT on POD 2 after insertion of a subduroperitoneal shunt showed complete regression of the bihemispheric subdural hygroma with the arachnoid cyst stable in size. Cranial T2- weighted MRI (E) and coronal T2 Flair section (F) at final routine examination 20 months after initial surgery shows the left-sided arachnoid cyst constant in size measuring $9 \times 7 \times 9$ cm. The bihemispheric subdural collections did not reoccur.

a burrhole reservoir, and a distal shunt catheter leading to the left peritoneal cavity, without the use of a pressure valve.

Postoperatively, the patient experienced significant relief from headache and visual disturbances, though a mild VI nerve palsy persisted. Radiological assessment on postoperative day 2 showed complete regression of the bihemispheric subdural hygroma, with the size of the temporosylvian arachnoid cyst remaining unchanged (Fig. 1 C/D). The patient was discharged on postoperative day 3 with complete resolution of headaches and markedly improved visual capabilities, achieving a Karnofsky Performance Score of 90 %. At the two-week follow-up, he had fully resumed his professional and daily activities without constraints. Persistent bilateral papilledema was noted, attributed to elevated ICP prior to surgery; however, it was expected to resolve over time. At the two-month follow-up, full visual acuity was restored, and papilledema had regressed on fundoscopy and optical coherence tomography (OCT). By four months postoperatively, papilledema had resolved, and visual field loss had disappeared within eight months.

At the ten-week follow-up, the patient reported no constraints (Karnofsky Performance Score 100 %), leading to the proposal for explantation of the SDP shunt device. With the hygroma resolved and a low likelihood of cyst rupture recurrence, the device was uneventfully removed three months post-surgery. Subsequent routine examinations at 6, 8, and 20 months confirmed stable conditions. MRI at the 20-month follow-up showed the left-sided arachnoid cyst unchanged in size, with no recurrence of the bihemispheric subdural collections (Fig. 1 A-F).

3.5.2. Case 2

A 22-year-old male patient presented emergently to our neurology team with a five-day history of severe left-sided frontotemporal headache associated with phonophobia, photophobia, nausea, and vomiting. The pain was described as stabbing and worsened with movement. The patient had a history of monthly recurrent headaches of similar quality but lesser severity, leading to an initial suspicion of status migrainosus. Treatment with triptans and metoclopramide provided relief, and the patient was scheduled for cranial imaging two days later.

A CT scan revealed an extra-axial, isodense, space-occupying lesion measuring $8.6 \times 4.4 \times 3.7$ cm on the left side, extending into the frontal region, and a subdural fluid collection (maximum 14 mm in width) along the left hemisphere. This was accompanied by a 15 mm rightward midline shift, uncal, and subfalcine herniation. These findings were confirmed by cranial MRI (Fig. 2 A/B). The diagnosis of a ruptured arachnoid cyst causing a subdural hematoma and elevated intracranial pressure (ICP) was established.

The patient was admitted to our neurosurgical department, presenting with severe headache that had recurred the previous night, accompanied by fever (Karnofsky Performance Score 80 %). Neurological examination was otherwise intact, and internal and biochemical investigations showed no significant abnormalities. The patient reported regular cannabis use and a family history of migraine; he worked full-time in real estate.

Urgent surgical intervention was required. We performed a burr hole trepanation on the left side to evacuate the subdural hematoma, followed by the implantation of an SDP shunt as previously described. The postoperative course was uneventful. The patient showed significant improvement and stability without focal neurological deficits. Postoperative MRI on day 1 and shunt series confirmed correct placement of the SDP shunt and significant regression of the parietal subdural hematoma. The arachnoid cyst remained stable in size, with resolution of hemorrhagic components, reduction in midline shift, and herniation (Fig. 2 C/D). Postoperative neuro-ophthalmological examination revealed bilateral papilledema, consistent with elevated ICP prior to surgery. The patient was discharged on postoperative day 3, clinically stable and asymptomatic (Karnofsky Performance Score 90 %).

Follow-up visits at one week and one month showed the patient to be asymptomatic, pain-free, and unrestricted in daily activities (Karnofsky Performance Score 100 %). Based on the favorable outcome at the three-

month postoperative follow-up, the shunt system was removed five months after the initial surgery. MRI performed five months postoperatively demonstrated complete resolution of the subdural hematoma, with the arachnoid cyst remaining stable in size (Fig. 2A-F).

Both patients exhibited immediate symptom regression following surgery and satisfactory long-term outcomes. Our experience indicates that while papilledema typically resolves over time, it may persist if ICP has been elevated for an extended period before intervention. After confirming the absence of recurrent intracranial fluid collections radiologically, the removal of the shunt system was deemed appropriate.

4. Discussion

While there is broad consensus that surgery is necessary for symptomatic cases following the rupture of an intracranial arachnoid cyst (IAC), the optimal surgical target and approach remain debated. The rarity of IAC rupture in young adults, coupled with the acute nature of its presentation, limits the feasibility of large prospective randomized controlled trials to determine the best treatment strategy among the various options, which differ significantly in terms of invasiveness and risk.

Our patients experienced an uneventful recovery and were able to quickly resume normal activities. Cranial MRI conducted shortly before and after shunt removal demonstrated that the IAC remained stable, with no residual or recurrent rupture or hemorrhage, and complete resolution of the previously identified subdural fluid collections. Long-term follow-up revealed no further complications. Our experience supports the efficacy and safety of subdural fluid drainage via burr hole trepanation combined with SDP shunt placement, without the use of interconnected shunt pressure valves. This approach, which allows for both immediate and gradual drainage of the subdural fluid collection, aligns with encouraging results from earlier series predominantly involving pediatric patients [6,14,18].

5. Conclusion

Our study highlights that this relatively minimally invasive technique is feasible for use in young adult patients, potentially avoiding more complex surgical methods that may pose unnecessary risks to otherwise asymptomatic individuals. We recommend considering this approach for similar cases in the future. However, further research with a larger sample size is needed to more comprehensively evaluate its efficacy and safety.

Ethical approval

Written informed consent was obtained from each patient in accordance with the local ethical requirements and the Declaration of Helsinki.

CRediT authorship contribution statement

Fassl Verena: Conceptualization, Methodology, Writing – original draft, Visualization, Investigation, Writing – review & editing. **Sebök Martina:** Investigation, Writing – review & editing. **Fierstra Jorn:** Investigation, Writing – review & editing. **Krayenbühl Niklaus:** Conceptualization, Investigation, Writing – review & editing. **Regli Luca:** Writing – review & editing. **Velz Julia:** Conceptualization, Methodology, Visualization, Investigation, Writing – review & editing.

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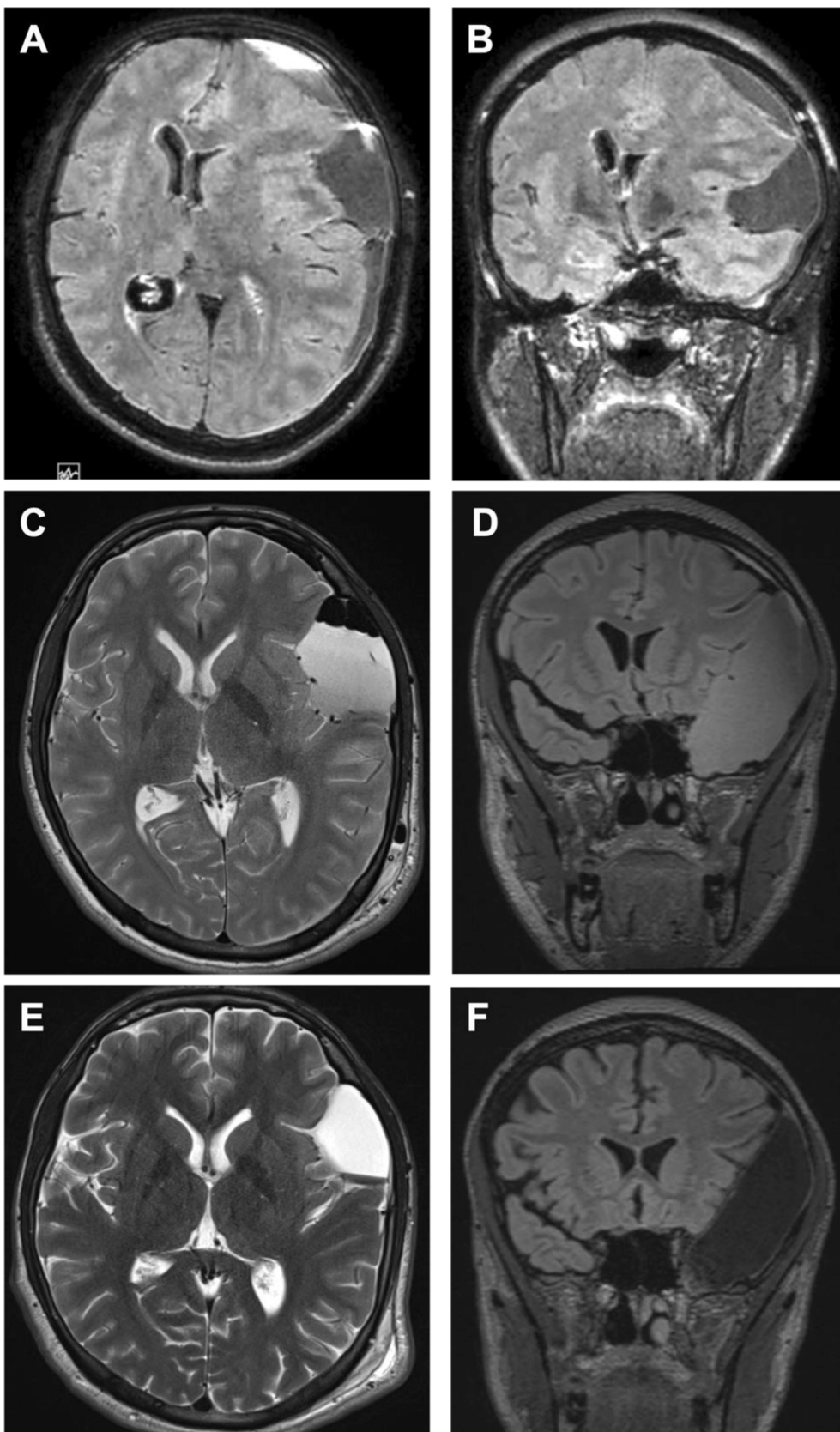


Fig. 2. Axial (A) and coronal (B) T2 FLAIR weighted MRI reveal a large, extraaxial, temporopolar CSF isointense space-occupying lesion measuring $8.6 \times 4.4 \times 3.7$ cm with left hemispheric subdural hematoma, resulting in contralateral midline shift of 12 mm, uncal and subfalcine herniation, and ballooning of the right lateral ventricle with transependymal edema. Axial T2-weighted (C) and coronal T2 Flair-weighted (D) MRI on POD 1 demonstrate significantly reduced subdural hematoma along the left hemisphere while the arachnoid cyst remained stable in size. Axial T2-weighted (E) and coronal T1-weighted (F) MRI 5 months postoperatively

show that the subdural hematoma did not reoccur. The arachnoid cyst, which extends from frontal to insular to temporopolar on the left side, remained constant in size.

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.

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