

# Infection of a Sylvian Galassi II arachnoid cyst after craniotomy for resection of a parasagittal meningioma

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## SUMMARY

Arachnoid cysts are CSF-containing entities that rarely are symptomatic or warrant neurosurgical intervention. In addition, infection of these lesions is an even rarer event, with only four reports in the literature capturing this. In this report, we present the case of a 79-year-old man presenting with paraparesis, secondary to a right parasagittal meningioma, with an incidental asymptomatic right Sylvian arachnoid cyst (Galassi type II). The initially planned surgery was postponed for 3 months, due to COVID-19 restrictions, and he was kept on high dose of steroids. Following tumour resection, the patient developed bilateral subdural empyemas with involvement of the arachnoid cyst, requiring bilateral craniotomies for evacuation of the empyemas and drainage of the arachnoid cyst. Suppuration of central nervous system arachnoid cysts is a very rare complication following cranial surgery with the main working hypotheses including direct inoculation from surrounding inflamed meninges or haematogenous spread secondary to systemic bacteraemia, potentiated by steroid-induced immunosuppression. Even though being a rarity, infection of arachnoid cysts should be considered in immunosuppressed patients in the presence of risk factors such as previous craniotomy.

## BACKGROUND

Arachnoid cysts (ACs) are developmental lesions lined with meningotheial cells that arise usually from splitting of the arachnoid membrane in early embryonal life. They contain CSF-like fluid and do not communicate with the ventricles or the subarachnoid spaces. They are found in 0.2%–1.7%<sup>1</sup> of the population and only around 5% of them become symptomatic, usually through local mass effect or due to adjacent brain tissue atrophy. More than 50% of them occur in the middle cranial fossa and show a predisposition for male gender and left side of the cranial vault.<sup>2</sup> They have also been linked with syndromic presentations such as Marfan's syndrome, NF1 and APKD.<sup>3 4</sup> Several hypotheses have been postulated in the pathogenesis of ACs. The strongest hypothesis supports the incomplete and aberrant separation of the pia-arachnoid layers at around the 15-week gestation point that leads to a loculated cystic lesion, in close proximity to an arachnoid cistern.<sup>5</sup> However, there is no consensus regarding the expansion of an AC, with the main hypotheses supporting active fluid secretion from the cyst walls,<sup>6</sup> an osmotic

gradient-driven expansion<sup>3</sup> or a slit-valve-like communication with the subarachnoid spaces.<sup>3 5</sup> In this report, we present the case of a 79-year-old patient who, following elective craniotomy for resection of a parafalcine meningioma, presented with bilateral subdural empyemas and suppuration within a previously asymptomatic and stable AC. To our knowledge, this has been the first report to highlight this rare phenomenon.

## CASE PRESENTATION

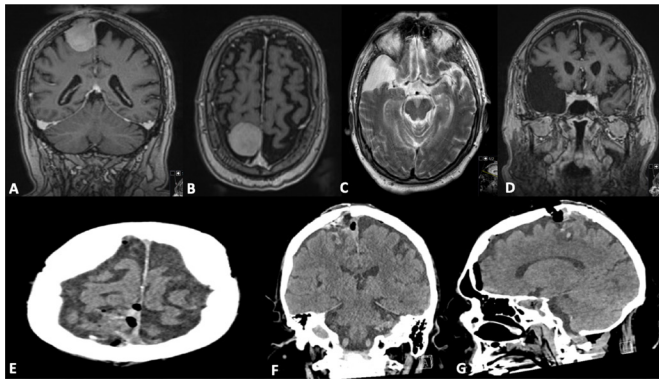
A 79-year-old man, with a significant medical history of pulmonary embolism and hypertension, presented to our neuro-oncology service with a 9-month history of progressive paraparesis and deteriorating balance with recurrent falls. Cranial imaging in the form of CT and MRI revealed a homogeneously enhancing right-sided parasagittal lesion consistent with a meningioma. There was also an incidental ipsilateral Galassi type II AC (figure 1A–D). Following discussion in the multidisciplinary meeting, the patient was commenced on an oral steroid regimen that led to improvement of his symptoms. Unfortunately, the patient required prolonged therapy with oral dexamethasone due to a 3-month delay in his operation date, due to COVID-19 restrictions. This was administered and weaned off as follows: day 0: 8 mg two times per day, day 6: 4 mg two times per day, day 10: 4 mg once a day, day 18: 2 mg once a day, day 84: 4 mg two times per day, with the surgery occurring on day 87. He underwent a right parietal craniotomy for resection of this lesion. The immediate postoperative period was uneventful and the patient recovered well with no complications or deficits and was discharged on the fifth postoperative day with a slow weaning dexamethasone regimen (half dose every 7 days to stopping the dexamethasone at the end. Postoperative imaging was satisfactory (figure 1E–G) and histopathological analysis revealed a WHO grade I meningioma.

He re-presented 7 days postdischarge, with complex partial seizures affecting his left upper limb, left upper limb monoparesis (MRC grade 4/5) and pustular discharge from his wound. Biochemical inflammatory markers were elevated while a CT brain precontrast and postcontrast did not demonstrate any deep tissue involvement. A diagnosis of surgical site infection was made (figure 2) and we, therefore, proceeded with a wound washout and removal of the bone flap. Surprisingly, however, the



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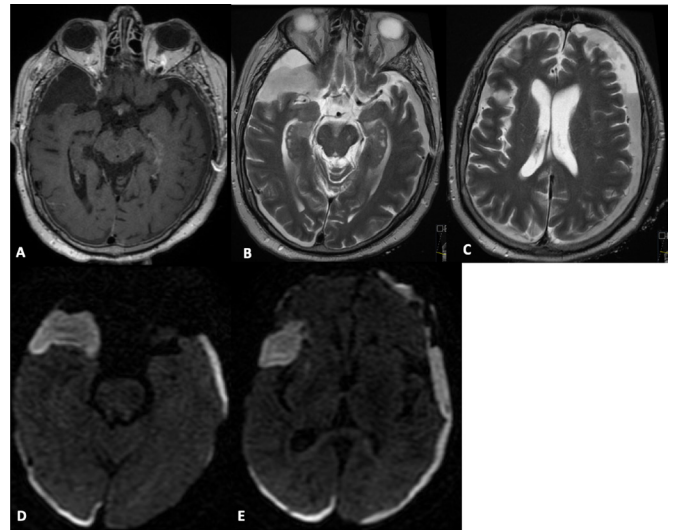
**Figure 1** Sagittal (A) and axial (B) T1W post-GAD MRI showing a uniformly enhancing right parasagittal lesion; axial (C) T2W MRI showing a right middle cranial fossa (Galassi type II) arachnoid cyst; coronal (D) T1W post-GAD MRI showing a non-enhancing right middle cranial fossa arachnoid cyst (Galassi type II); axial (E), coronal (F) and sagittal (G) postoperative plain CT brain. GAD, gadolinium; T1W, T1-weighted; T2W, T2-weighted.

CT brain revealed a left shallow, non-enhancing subdural collection, which was not in-keeping with the preservation of the dura intraoperatively. Despite its unclear and atypical occurrence, potential causes include: (1) venous spread across channels in the superior sagittal sinus, on a background of a pre-existing postoperative hygroma, (2) CSF spread via communication between the subdural spaces bilaterally; (3) arterial spread, even this is unlikely due to this not following a particular perfusion territory, and (4) direct inoculation, even this is unlikely, as the craniotomy was restricted to the right side, as per [figure 1](#).<sup>7</sup> Due to its non-enhancing nature, the lack of any underlying mass effect or any evidence of parenchymal irritation, the decision was made not to drain it, as this would essentially prevent seeding of infection to a potentially sterile collection. Intraoperatively, both epidural and subdural empyemas over the surgical site were identified and drained. Microbiological analysis revealed *Staphylococcus aureus* sensitive to flucloxacillin. He was, therefore, commenced on intravenous antibiotics, while, as per endocrinological advice, dexamethasone wean continued with added 50 mg of intravenous hydrocortisone, which was discontinued on postoperative day 21.

Unfortunately, he subsequently developed status epilepticus requiring intubation and sedation for seizure control. This was most likely due to overlying bilateral enhancing convexity collections with restricted diffusion, representing subdural empyemas, as demonstrate on a repeat MRI brain. The right AC was also involved, which was indicative of cyst suppuration ([figure 3](#)). Other parameters that could act as epileptiform triggers, such as biochemical derangement, hypoxia and pharmacological causes



**Figure 2** Axial (A), coronal (B) and sagittal (C) slices of the postoperative contrast-enhanced CT brain showing a left, non-enhancing subdural collection.



**Figure 3** Axial T1W MRI post-GAD (A), T2W (B, C) and diffusion-weighted images (D, E) demonstrating an enhancing periphery of the right arachnoid cyst, with associated fluid levels with this cyst and over the convexity as well restricted diffusion within these areas. This indicated subdural empyema and suppuration of the arachnoid cyst. Please note that bilateral hippocampal region hyperintensities were also present on preoperative imaging and do not represent an acute event. GAD, gadolinium; T1W, T1-weighted; T2W, T2-weighted.

were ruled out. He, therefore, underwent bilateral craniotomies to evacuate the subdural empyemas and to also drain the AC.

#### OUTCOME AND FOLLOW-UP

Despite his good immediate postoperative neurological recovery, he developed hospital-acquired pneumonia secondary to *Pneumocystis jirovecii* and died shortly after.

#### DISCUSSION

ACs are usually incidental radiological lesions that follow a benign clinical course. The strongest indication for imaging the brain include trauma and persistent neurological symptoms or headaches. They are more prevalent in the paediatric population and show a predisposition for the middle cranial fossa. Only a small percentage of 5% become symptomatic, usually due to temporal enlargement of the cyst. They usually manifest with symptoms of raised intracranial pressure, such as headaches, cognitive deficits, ataxia, vertigo, hearing loss, seizures, while developmental delay and progressive macrocephaly is more evident in the paediatric population.<sup>8</sup> Middle fossa ACs have been associated more with headaches, seizures and motor deficits,<sup>3</sup> while more pronounced local effect has been reported to lead to temporal lobe agenesis.<sup>9</sup>

Generally, infection of such lesions is a very rare phenomenon, with only two adult and one paediatric cases reported in the literature ([table 1](#)).<sup>10–12</sup> The above cases report spontaneous infections of ACs in preoperative patients who were not immunosuppressed. Our report has been the first to describe a postoperative infective complication of an AC. Our working hypothesis suggests that the subdural infection and subsequent meningeal inflammation resulted in the direct inoculation of the cyst wall. Another hypothesis involves haematogenous spread and systemic bacteraemia through the local draining channels directly to the AC. Intraoperative appearances demonstrated a macroscopically inflamed cyst wall with a pustular collection

**Table 1** Summary of the two adult case reports, highlighting infected ACs

Case	Authors	County	Year	Age/sex	Background	Symptoms	Imaging	Surgery	Organism	Outcome
1	Park <i>et al</i> <sup>10</sup>	Korea	2013	53 Female	Chronic sinusitis	5 months of headaches with 1 month of hormonal imbalances (diabetes insipidus, dysmenorrhoea)	Sellar cystic mass with thickened pituitary stalk	Transphenoidal drainage of the cyst	Not disclosed	Collapse of the cyst with ongoing vasopressin due to ongoing diabetes insipidus
2	Sivaraman <i>et al</i> <sup>11</sup>	UK	2007	83 Female	Previous stroke with mild residual hemiparesis atrial fibrillation	Pneumonia followed by left hemiparesis and seizures	Right frontal arachnoid cyst with areas of restricted diffusion	Craniotomy for drainage of the cyst and extirpation of the cyst wall	<i>Streptococcus pneumoniae</i>	No further seizures Recovered to left sided MRC 5/5 power
3	Gale <i>et al</i> <sup>12</sup>	USA	2020	7 Male	Chronic hearing loss	Headache, fever, difficult ambulation	Sinusitis Left temporal AC with subsequent leptomeningeal enhancement and abscess formation within the AC	Left craniotomy with cyst fenestration and washout	<i>S. pneumoniae</i>	Right facial droop Right hemiparesis Complete bilateral sensorineural hearing loss

ACs, arachnoid cysts.

residing in its cavity. The long-term symptomatic therapy with corticosteroids the patient was receiving in the interim between the decision to operate and the operation date was likely to have contributed to and created an immunodeficient state that increased his vulnerability to infections. This was indeed one of the negative consequences the first wave of the COVID-19 pandemic instigated, with mandatory delays and cancellations in elective surgeries, that translated into a 3-month unnecessary corticosteroid therapy in this patient.<sup>13</sup> As a consequence, this patient developed opportunistic infections which eventually led to septicaemia and despite the initially excellent neurological recovery, the patient's death.

This case has allowed us to reflect on our oncology practice in patients with incidental ACs, as the risk of potential post-operative infection is not one to be underestimated. Patients undergoing elective craniotomies with incidental ACs should be counselled on potential complications, such as subsequent infection of the AC. In addition, we should highlight again the negative consequences of long-term immunosuppression in benign brain tumours, which might impact on the patient's immune competence, a practice well documented in the literature. Last but not least, we should highlight the impact COVID-19 pandemic has had on the neurosurgical community, with services being run in obstructive and suboptimal ways, which could essentially lead to unfavourable outcomes.

### Patient's perspective

Patient is deceased.

### Learning points

- ▶ Traditionally benign or incidental intracranial pathologies should be considered as a potential cause of morbidity and mortality, in a patient undergoing cranial surgery.
- ▶ Patients on long-term immunosuppressive therapy should be appropriately counselled for potential postoperative complications.
- ▶ COVID-19 has had a significant impact on neurosurgical practice.

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