

A Case with Ruptured Arachnoid Cyst Complicated with Spontaneous Intracranial Hypotension

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ABSTRACT Arachnoid cysts are mostly benign, asymptomatic lesions but serious complications for instance subdural haematoma, intracystic haemorrhage and subdural effusion following rupture of the cyst are seen infrequently. We present a novel case with a ruptured temporal arachnoid cyst, which was complicated by the existence of coincidental and confusing clinical and radiological findings of spontaneous intracranial hypotension. Displaying the link between the right temporal arachnoid cyst and subdural effusion in the cranial magnetic resonance imaging (a flow-sensitive 3D section T2 space sequence) played a critical role in our diagnosis. Aim of this presentation is to discuss our treatment and management approaches in a complicated case of ruptured temporal arachnoid cyst and emphasize on an appropriate MR sequence for a diagnosis.

Keywords: Arachnoid cysts; intracranial hypotension; intracranial hypertension; subdural effusion; cerebrospinal fluid leak

Arachnoid cysts are mostly benign, asymptomatic lesions but they may be related to complications such as subdural hematoma, subdural hygroma, and intracystic hemorrhage. Minor head injury might result in subdural hygroma or hematoma due to rupture of arachnoid cyst.¹

Spontaneous intracranial hypotension can be a rare but curable cause of severe and restrictive headaches. Typically, the headache is orthostatic, worsening when the sufferer assumes an upright position and improving when the person lies down. The clinical symptoms of spontaneous intracranial hypotension may vary because of traction on structures of the central nervous system (such as cranial nerves or spinal roots) or prolonged venous engorgement due to low cerebrospinal fluid (CSF) pressure. Neuroimaging is quite helpful for secondary headache diagnosis. In spontaneous intracranial hypotension, subdural effusion, pachymeningeal thickening, engorgement of venous structures, pituitary hyperemia and sagging of the brain can be discerned on magnetic resonance imaging (MRI). However, the spectrum of clinical and radiological findings is broad and diagnostic failure is common. We present a novel case with a ruptured temporal arachnoid cyst, which was complicated by the existence of coincidental and confusing clinical and radiological findings of spontaneous intracranial hypotension.

CASE REPORT

A 24-year-old woman was hospitalized in a foreign country complaining of an orthostatic headache that had started twenty days prior. She had been treated with a diuretic therapy for 5-days. Her neurological examination was normal before the diuretic medication, then the headache worsened and diplopia ensued. After her clinical deterioration, the patient came to our hospital. She had suffered a minor head trauma a month before, but had no complaints other than constipation. Bilateral mild papillary edema and bilateral abducens nerve palsies, predominantly on the left, were detected in her neurological examination. Bilateral subdural effusion, diffuse dural enhancement (Figure 1 A, B), a temporal arachnoid cyst on the right side and mega cisterna magna (Figure 1 C, D) were noted on the contrast-enhanced cranial MRI. Magnetic res-

onance neurography showed CSF leaks at L3-L4, L4-L5 distances on the left side (Figure 2 A). An injection of autologous blood in the lumbar epidural space was performed (20 mL). Two days after the treatment the patient had made no improvement and once again the autologous epidural blood patch was repeated (20 mL). After the second procedure, the headache was markedly relieved and the patient was discharged without neurological pathology except for the mild sixth cranial nerve paralysis on the left side, with the recommendation of copious hydration and caffeine.

Two weeks later, she returned with continuous headache and complained of double vision. Her neurological examination showed bilateral papillary edema (grade 4) and severe bilateral abducens deficit mostly affecting the left eye. In the cranial MRI, the left subdural effusion was almost restored, the right subdural effusion had become more evi-

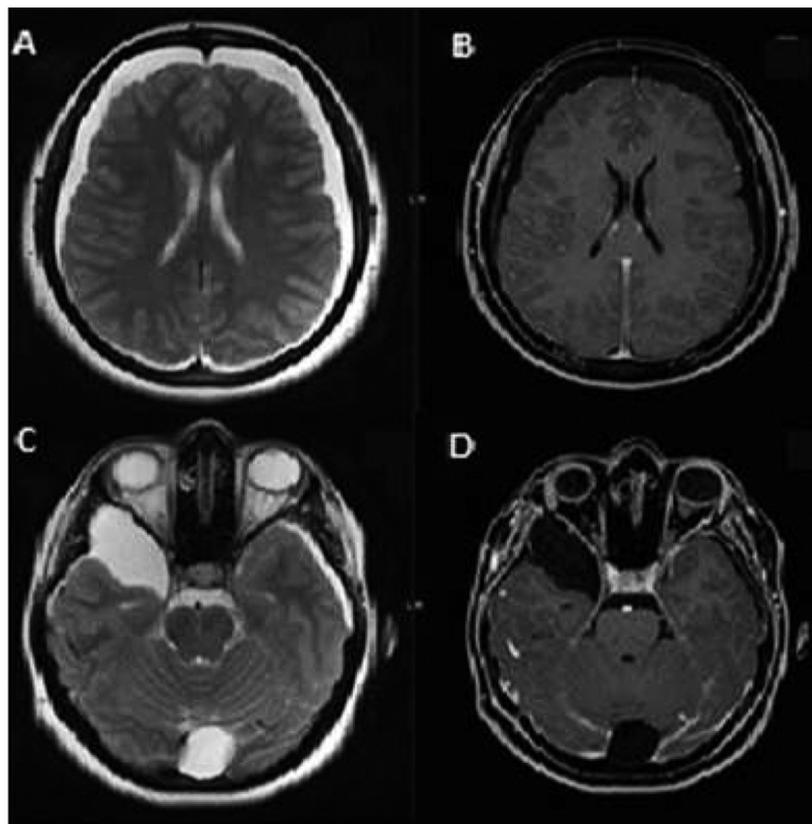


FIGURE 1: Bilateral subdural collection (A) and dural contrast enhancement (B) are observed on T2 (A) and contrast-enhanced T1-weighted (B) axial cranial MR images. Dural contrast enhancement in part B was indicated for an arrow. T2 (C) and T1 (D) axial cranial MR images show the arachnoid cyst on the right temporal pole and mega cisterna magna.

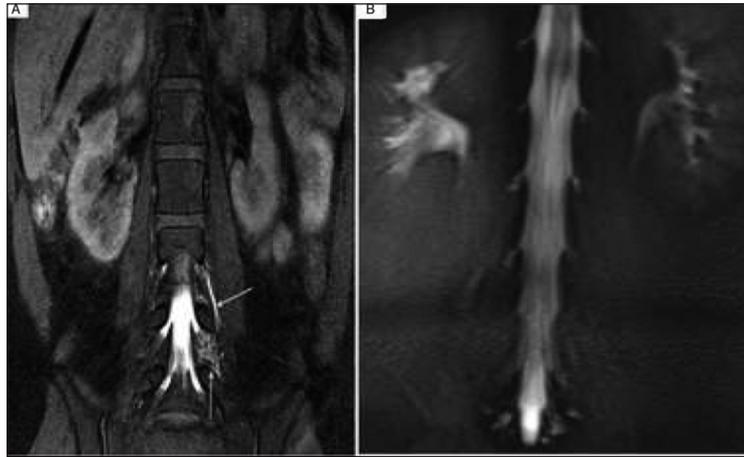


FIGURE 2: (A) Intrathecal gadolinium injected T1-weighted coronal image on MR neurography shows CSF leaks at L3 and L4 roots in the left foramina (arrows). **(B)** Intrathecal gadolinium injected T1-weighted coronal image on MR neurography does not demonstrate any leak.

dent and the midline structures had shifted to the left side (Figure 3 A). The possibility of venous sinus thrombosis was investigated because of the rebound headache and intracranial hypertension findings.² Intracranial MR venography was found normal with no evidence of sinus thrombosis. Contrast-enhanced MRI of the lumbar spine did not show any leak (Figure 2 B). Given the worsening neurological (intractable headache, cranial nerve paralysis, papillary edema) and radiological findings, an epidural blood patch (20 mL) was performed once again. As noninvasive treatment was unable to control clinical and radiological deterioration, burr hole surgery to drain the subdural effusion on the right side was performed to help relieve the pressure on the brain due to intracranial hypertension.

On her cranial MRI, a flow-sensitive 3D section T2 space sequence showed hypo-intense flow compatible with communication between the right temporal arachnoid cyst and the subdural effusion (Figure 3 B). Based on the radiological findings, a right fronto-temporo-parietal craniotomy and surgical treatment of the arachnoid cyst were performed by the neurosurgeon. Regression of the midline shift of the brain was observed on radiological follow-up. One week after the surgery, significant improvement was reported in her headache and adduction deficit of the eyes. The patient has been headache-free since for nearly one month and the radiological findings have shown

major improvement (Figure 4). Since the patient lives abroad, she obtained an MRI at another hospital which she shared with us via email. The image revealed almost complete resolution for all radiological findings after three months.

DISCUSSION

Arachnoid cysts are the most common congenital cystic abnormality in the brain. They are usually found incidentally. In general the course of arachnoid cysts is uneventful. Grave complications such as subdural haematoma, intracystic haemorrhage and subdural effusion following rupture of the cyst are seen infrequently.^{3,4} The relation between arachnoid cysts and subdural hygromas is considered exceptionally rare and only a few cases have previously been reported.³ There are two likely mechanisms for the pathogenesis of subdural hygromas. The first is a linking between the cyst and the subarachnoid space owing to a minor head injury with a flap-valve mechanism so that there is a flow of CSF from the subarachnoid space into the cyst. The flap-valve mechanism causes an increase in the size and pressure of the cyst leading to a rupture of the cyst into the subdural space.⁴ Another mechanism is a sudden temporary raise in intracerebral pressure causing a rupture of the cyst wall into the subdural space during a Valsalva maneuver.^{4,5}

Spontaneous intracranial hypotension has been recognized as a curable and under-diagnosed

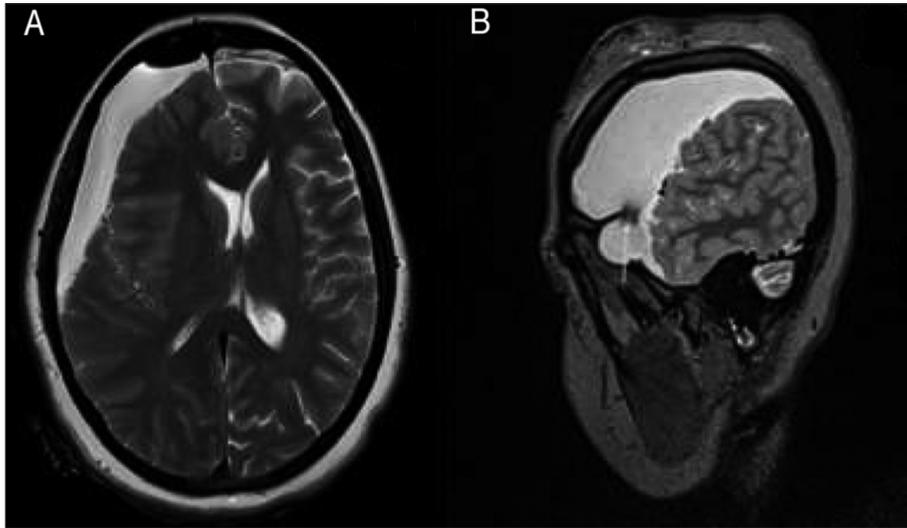


FIGURE 3: T2 weighted image (A) shows that the right subdural effusion had become more evident and the midline structures had shifted to the left side. A subdural hemorrhage of the left frontal can also be seen. A flow-sensitive 3D section T2 space sequence (B) illustrates hypo-intense flow compatible with communication between the right temporal arachnoid cyst and the subdural effusion.

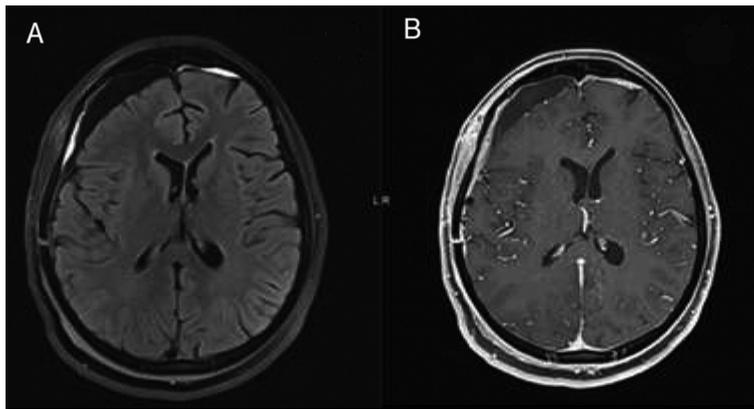


FIGURE 4: Flair (A) and contrast-enhanced T1-weighted (B) axial cranial MR images demonstrate the regression of the midline shift and subdural effusion of the brain at one-month radiological follow-up.

secondary cause of headache. Most cases of spontaneous intracranial hypotension are due to the spontaneous slow leakage of CSF via small dural defects. The exact mechanism of these dural defects is unknown, but it is thought to indicate the underlying meningeal weakness. This causes a sagging of the brain, traction of the bridging veins, and pain-sensitive meningeal structures and cranial nerves. According to the Monroe-Kellie hypothesis, secondary venodilation may develop as a compensatory measure to the low CSF pressure. Adding a vascular component with venous dilatation could represent SEEPS (subdural hygroma, enhancement,

engorged veins, pituitary hyperemia, and sagging of the brain) on cranial MRI.

Before the patient came to our hospital, she was misdiagnosed and treated with diuretics. In our hospital, we made a diagnosis of spontaneous intracranial hypotension because of orthostatic headache and MR myelography that demonstrated CSF leaks at L3-L4, L4-L5 distances on the left side. So both conservative and invasive approaches for treatment of spontaneous intracranial hypotension were applied but her neurological condition got worse. Then the MRI showed a hypo-intense flow compatible with communication between the right temporal

arachnoid cyst and the subdural effusion, we finally thought that increased intracranial pressure is probably associated with arachnoid cyst rupture. After surgical treatment of the arachnoid cyst, the patient improved. Our first diagnosis for spontaneous intracranial hypotension was made based on MR neurography that showed CSF leaks. Displaying the link between the right temporal arachnoid cyst and subdural effusion on MRI played a critical role in the treatment plan. A flow-sensitive 3D section T2 space sequence was finalized our diagnosis.

The purpose of this presentation was to discuss our treatment and management approaches in a complicated case of ruptured temporal arachnoid cyst and emphasize on an appropriate MR sequence for a diagnosis. We hope this will provide additional insights into this neglected area of medical knowledge.

Informed Consent

Patient gave written informed consent before the study, and the paper was approved by the human ethics committee of the hospital.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Seda Kosak; **Design:** Pınar Yalınay Dikmen; **Control/Supervision:** Pınar Yalınay Dikmen, Elif Ilgaz Aydınlar; **Data Collection and/or Processing:** Ercan Karaarslan, Mehmet Zafer Berkman; **Analysis and/or Interpretation:** Pınar Yalınay Dikmen, Seda Kosak; **Literature Review:** Seda Kosak; **Writing the Article:** Seda Kosak, Pınar Yalınay Dikmen; **Critical Review:** Mehmet Zafer Berkman.

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