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Case report

Spontaneous subarachnoid hemorrhage due to arteriovenous malformation mimicking migraine: A case report ☆

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ABSTRACT

Subarachnoid hemorrhage (SAH) due to Arteriovenous Malformation (AVM) is a rare emergency case, which is often misdiagnosed as migraine. Here we present a case of SAH due to AVM that mimics migraine. A 41-year-old man came with headaches that radiated to the neck, worsened in the last week, accompanied by nausea, vomiting, photophobia, and a history of intermittent headaches for the previous 2 years. Physical examination was within normal limits, initial laboratory tests showed leukocytosis, and CT scan was not typical. The patient was diagnosed with migraine. Apparently, the lumbar puncture showed very high red blood cells, suspected as SAH. CT angiography revealed an extra-axial AVM. The patient was later diagnosed as SAH due to AVM. We recommend applying 4 key points, namely headache progressivity, neck pain, neck stiffness, and leukocytosis, to differentiate SAH due to AVM from migraine, especially in areas with limited facilities.

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Introduction

Subarachnoid hemorrhage (SAH) is bleeding in the subarachnoid space, generally caused by trauma (80%) [1]. SAH is a neurological emergency, with mortality rates ranging from 8%-67% [1]. This high mortality makes identifying the underlying cause of SAH crucial, primarily to aim for fast initial treatment.

Arteriovenous malformation (AVM) is one of the causes of SAH that is rare but very important to be identified and treated immediately, mainly because AVM rupture has a risk of rebleeding up to 20% [2]. AVM is an abnormal blood vessel (nidus) that drains blood directly from arteries to veins without passing through the capillary system [2]. Because of the high risk of rebleeding and the rapid emergency progression, identifying patients with AVM-associated SAH is essential, significantly to differentiate them from other diseases with similar symptoms.

Here, we report a case of spontaneous SAH due to AVM that mimics migraine symptoms. Here we try to explain our diagnosis approach and provide critical key points that could help distinguish these 2 diseases so that spontaneous SAH could be diagnosed and managed quickly, especially in areas with limited facilities.

Case illustration

A 41-year-old man came with complaints of severe headache 1 week before being admitted to the hospital. Throbbing pain on both sides of the head radiating to the neck with Numerical Rating Scale (NRS) 8, frequency every 2-3 months, improves when in a dark and quiet place, and does not get worse with coughing. Headaches have been felt for the previous 2 years but have gotten worse in the last week. There is nausea and vomiting more than 5 times per day. There were no other associated symptoms such as an aura, loss of consciousness, behavioral changes, fever, side weakness, or seizures. The patient has no history of head trauma, stroke, or chronic disease. The patient has already received the third COVID-19 vaccine and his COVID-19 PCR test was negative.

On physical examination, vital signs were normal, Glasgow Coma Scale 15, no meningeal signs, and no neurological deficit. The patient's laboratory test results at the time of admission showed a leukocytosis the remainder were normal.

The patient's initial working diagnosis was migraine headache based on the history, physical examination, and initial investigations. Head computed tomography (CT) scan with contrast was performed within the first 1 hour and showed symmetrical dilatation of the lateral and third ventricles without signs of obstruction, meningeal enhancement, mass in the brain, infarction, or hemorrhage (Figs. 1A-C)

On the second hospital day, a lumbar puncture was performed, as shown in Figure 2A. Lumbar puncture examination revealed an opening pressure of 38 cmH2O, reddish cerebrospinal fluid, red blood cell counts 9000 cells/μL, increased cell count, increased cerebrospinal fluid protein, and decreased cerebrospinal fluid glucose, suggesting infection. Although the lumbar puncture results showed an infection,

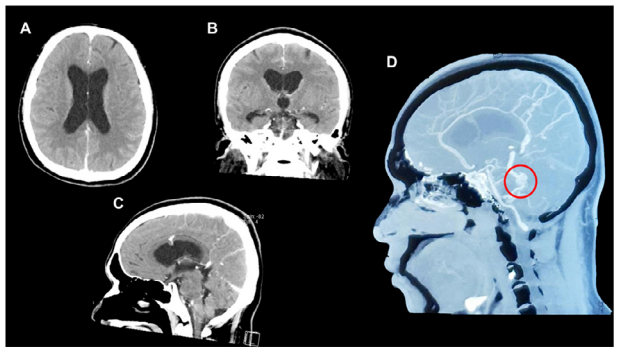

Fig. 1 – Head CT scan head with contrast on (A) axial, (B) coronal, and (C) sagittal sections. Cerebral CTA examination (D) was also performed, showing an extra-axial AVM with a 1.2 cm nidus on the anterior wall of the fourth ventricle (red circle).

the sign of redness and a very high number of red blood cells in the cerebrospinal fluid should be suspected as SAH. Even 3 CSF tubes showed a persistent red color (Fig. 2B). Based on this result, cerebral CT angiography (CTA) with contrast was performed on the third hospital day. The CTA shows an extra-axial AVM with nidus measuring 1.2 cm in the fourth ventricle's anterior wall, which received a feeding artery from the basilar artery and draining vein to the left Rosenthal basal vein (Fig. 1D). There was no intracranial aneurysm, intracranial hemorrhage, infarction, or space-occupying lesions.

Based on the history, physical examination, and all investigations, the patient's working diagnosis was SAH (Hunt-Hess 1, modified Fisher 0) caused by extra-axial AVM rupture (Spetzler-Martin 3). The patient was given oral nimodipine 60 mg q.6.d., intravenous ketorolac 30 mg t.i.d., morphine 1mg if accompanied by severe headache, and planned embolization.

Until the fourth hospital day, the patient still complained of headache with NRS 8, so the patient's therapy was added with oral paracetamol 1000 mg t.i.d. On the fifth hospital day, the patient's headache reduced with NRS 6 but was still accompanied by vomiting 3 times per day. Intravenous tramadol 50 mg b.i.d. and intravenous ondansetron 8 mg b.i.d. were added. On the sixth hospital day, the patient's headache was reduced with NRS 5, still accompanied by vomiting 4 times per day, and the patient's tramadol dose was increased to 50 mg t.i.d. On the eighth hospital dayt, the patient's headache decreased with NRS 3 and no vomiting. The patient's headache decreased on the ninth hospital day with NRS 2, and on the tenth hospital day, the patient was no longer complaining of a headache. The patient was discharged at his request on the eleventh hospital daywith an outpatient embolization plan.

Discussion

Diagnosing SAH is crucial to providing appropriate treatment in an emergency setting. SAH is sometimes associated with a very severe thunderclap headache, which occurs suddenly. However, in this case, SAH due to AVM showed progressive

A

CSF Analysis	Normal Range	Second Day of Treatment
Color	Colorless	**Redness**
Clarity	Clear	**A bit cloudy**
Blood clot	Negative	Negative
Cytology (Cell type)	0-5 cell/uL	**9000 cell/uL (RBC)**
Indian ink	No cryptococci found	No cryptococci found
Protein	14-45 mg/dL	**55 mg/dL**
Glucose	50-80 mg/dL	**43 mg/dL**
Serum Glucose	60-140 mg/dL	*serum not included
Chloride	115-130 mg/dL	121 mg/dL
Gram staining	No bacteria found	No bacteria found
AFB stain	Negative	Negative
PCR TB	Negative	Negative

AFB: Acid Fast Bacilli; **CSF:** Cerebrospinal Fluid; **PCR:** polymerase chain reaction; **RBC:** red blood cells; **TB:** tuberculosis

B

Fig. 2 – Analysis of the patient's cerebrospinal fluid. (A) Laboratory Analysis. (B) CSF with persistent red color in all 3 tubes.

and atypical symptoms, even mimicking migraines, making the diagnosis challenging. This section will describe our diagnostic approach and key points in differentiating atypical spontaneous SAH with migraine.

The key points that need to be identified from history taking are the progression of the headache and the presence of neck pain. In our case, the patient came with NRS 8 headaches in both heads that radiated to the neck and got worse in the last week. Although the headache has great intensity, there has been a history of intermittent headaches since 2 years ago and has only worsened in the last week. The duration and onset of these headaches do not match the typical headache of SAH, namely thunderclap headache so that the differential diagnosis of migraine is still possible. However, the 2-year course of severe headache may be due to slowly progressive AVM due to high blood flow supply from the basilar artery [3]. Furthermore, the worsening headache in the past week may be due to the gradual rupture of the AVM, thus avoiding a thunderclap headache. Sugita et al. [4] also reported a similar clinical picture of gradual onset of headache in an 11-year-old AVM patient. In addition to progression, neck pain can also be a clinical symptom that supports the diagnosis of SAH. A meta-analysis by Carpenter et al. [5] showed that the history of neck pain (positive likelihood ratio [LR+] = 4.1) significantly increased the likelihood of SAH. In addition, the presence of photophobia and phonophobia in these patients is also not specific for migraines since SAH with AVM could have similar symptoms [6].

In addition to history taking, the physical examination also plays an important role in differentiating SAH from migraine. Carpenter et al. [5] showed that neck stiffness (LR+ = 6.6) significantly increased the likelihood of SAH. However, there was no abnormality on physical examination in our patient, which may be based on slowly progressive SAH due to AVM. Patients suspected of SAH with normal physical examination are also frequently found by clinicians [7]. This makes 12%-51% of patients with SAH initially misdiagnosed, most often diagnosed as migraine or tension-type headache [7].

The key point in the initial laboratory test in differentiating SAH from migraine is the presence of leukocytosis. Söderholm et al. found that leukocytosis was associated with increased incidence of SAH [8]. On the other hand, there is no evidence showing an association between migraine and leukocytosis. Nonetheless, the study of Karabulut et al. [9] showed that an increase in the neutrophils/lymphocytes ratio was associated with migraines. However, in our patients, the neutrophils/lymphocytes ratio was in the normal range and thus did not support the diagnosis of migraine.

In areas with adequate health facilities, the diagnosis could be followed by a CT scan, CSF analysis, or CTA. In our case, the patient underwent a CT scan of the head with contrast based on the Ottawa SAH rule, namely because the patient's age was more than or equal to 40 years. On CT scan of the head with contrast, the patient did not find any bleeding, even though the patient had SAH. This is most likely due to the patient's 7-day onset of headache, so the CT scan sensitivity is also reduced to only 50%-60% [10].

Because the head CT scan with contrast was non-specific, the patient was examined for a lumbar puncture. The results of a lumbar puncture with reddish CSF up to the third bottle and a red blood cell count of 9000 suggest SAH. This is supported by the study of Carpenter et al. [5] which indicated that the RBC count at a threshold of 1000/μL had a role in the diagnosis of SAH with LR+ was 5.7 [1.4-23] and LR− was 0.21 [0.03-1.7]. Based on the patient's LP results suggestive of SAH, the patient's working diagnosis changed to spontaneous SAH. CTA was performed as first-line vascular imaging, and a nidus was obtained, which ultimately confirmed the diagnosis of SAH due to AVM.

Conclusion

SAH due to AVM is a rare emergency case, which is often misdiagnosed as a migraine. The clinician's role in conducting history taking, physical examination, and initial laboratory testing is crucial. We recommend applying 4 key points, namely headache progressivity, neck pain, neck stiffness, and leukocytosis, to differentiate SAH due to AVM from migraine. These key points are especially useful in limited facilities, such as no access to CT scans, CTAs, or advanced laboratory tests.

Authors' contributions

Conceptualization, A.R., R.M., S.W.N., and S.Y.; methodology, A.R., R.M., S.W.N., and S.Y.; software, S.Y., E.W.; validation, A.R., T.M., M.K., R.H., R.M., S.W.N., and S.H.; formal analysis, S.Y.; investigation, A.R. and S.Y.; resources, A.R., T.M., M.K., R.H., R.M., S.W.N., and S.H.;data curation, A.R., T.M., M.K., R.H., R.M., S.W.N., and S.H.; writing—original draft preparation, S.Y. and E.W.; writing—review and editing, A.R.; visualization, R.M. and S.Y.; supervision, A.R., T.M., M.K., R.H., R.M., S.W.N., and S.H.; project administration, A.R.; funding acquisition, A.R., T.M., M.K., R.H., R.M., S.W.N., and S.H. All authors have read and agreed to the published version of the manuscript.

Patient consent

Informed consent was obtained from all subjects involved in the study.

Data availability statement

No new data were created or analyzed in this study. Data sharing is not applicable to this article.

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