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Case Reports & Case Series

A case of multiple infectious intracranial aneurysms concurrently presenting with intracerebral hemorrhage and epistaxis

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ABSTRACT

Infectious intracranial aneurysms (IIAs) have the specific features of rapid growth and multiplicity, which often show various pathologies and serious symptoms when ruptured. There are few previous reports of multiple IIAs that concurrently present with intracerebral hemorrhage (ICH) and epistaxis. A 62-year-old man with a history of tooth extraction 1.5 months before experienced an intracranial abscess in the left Sylvian fissure and insula cortex. The patient underwent a craniotomy to drain the abscess and he experienced a heavy epistaxis, which required intranasal gauze packing immediately after surgery. Subsequently, he had a massive ICH on the left temporal lobe resulting from a ruptured IIA in the left middle cerebral artery on day 8. Surgical trapping of the aneurysm contributed to a complete cure. Oral indigenous bacteria was obtained by culturing an aspirate of the abscess cavity. A follow-up radiological examination revealed that the right internal carotid artery (ICA) aneurysm in the cavernous segment concurrently grew and the aneurysmal rupture had caused the epistaxis. We performed stent-assisted coil embolization for the aneurysm on day 20 and achieved radiological resolution. Although right hemiparesis and some communication problems remained, the patient recovered but needed minimal assistance. Given multiple progressions and their destructive pathology, more attention needs to be paid to IIAs. Our multimodal strategy achieved clinical resolution, and IIAs should be treated immediately with appropriate radical intervention depending on the lesion location.

1. Introduction

Infectious intracranial aneurysm (IIA), which accounts for 1–5% of all intracranial aneurysms, is a distinctly rare cause of intracranial subarachnoid hemorrhage (SAH) [1]. This pathology frequently results from infectious endocarditis that is spread through the blood. There are various causative diseases such as bacterial meningitis, cavernous sinus phlebitis, or poor dental hygiene, which can lead to direct extravascular invasion. IIAs that are small in size or have a distal location may sometimes disappear after antibiotic treatment [2]. However, patients with severe IIAs often show serious pathology, with a mortality rate of more than 80%. Recent endovascular therapy has achieved good results as a less invasive intervention [3].

Although the IIA often has exhibited multiple lesions, previous publications on multiple ruptured IIAs are limited. We report an experience with multiple IIAs that concurrently presented with intracranial hemorrhage (ICH) and epistaxis. Surgical and endovascular procedures contributed to the radical cure. Diagnostic and technical management of the patient is also discussed.

2. Case presentation

A 62-year-old man with a history of tooth extraction 1.5 months before noticed diplopia and pain in the back of his right eye, and he visited another clinic 24 days before presenting to our hospital. He had taken oral medication for hypertension and diabetes for 2 years. Although his headache had developed gradually, abnormal findings on magnetic resonance image (MRI) and magnetic resonance angiography (MRA) were not apparent at his first and 18-day follow-up visits at the other clinic (Fig. 1). He was referred to our hospital because of

Abbreviations: IIA, infectious intracranial aneurysm; SAH, subarachnoid hemorrhage; ICH, intracerebral hemorrhage; MRI, magnetic resonance imaging; CT, computed tomography; DW-MRI, Diffusion weighted-MRI; CTA, CT angiogram; MCA, middle cerebral artery; ICA, internal carotid artery; MRA, MR angiography

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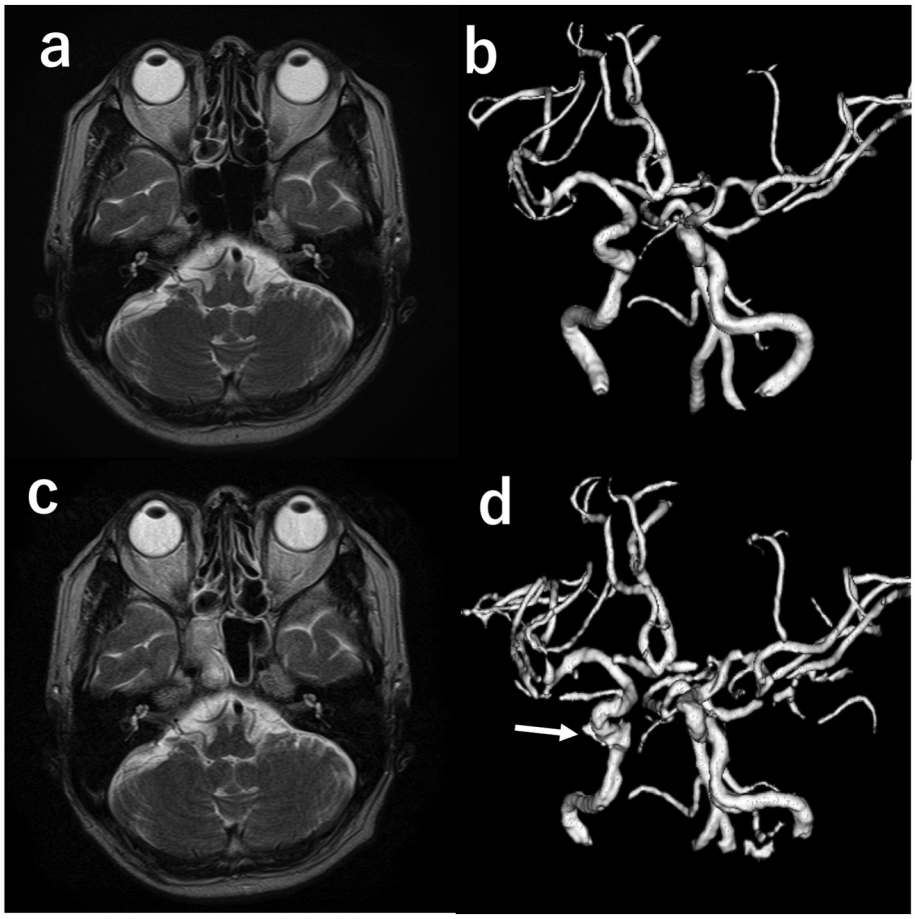

Fig. 1. (a), (b) T2-weigted magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) performed at our patient's first visit to the previous clinic showed no apparent abnormal findings. (c), (d) Follow-up images 6 days before admission to our hospital showed fluid collection in the right sphenoid sinus and an irregular-shaped vessel at the cavernous segment of the right internal carotid artery (d, arrow).

consciousness disturbance and status epilepticus.

At our institution, neurologic deficits with right facial palsy and right hemiparesis were observed. His consciousness level was drowsy according to the Glasgow coma scale (E4V1M6), and his pupils were reactive. Diffusion weighted-MRI (DW-MRI) showed hyper-intense lesions at the left insula cortex and subarachnoid space adjacent to the Sylvian fissure (Fig. 2a). T1-weighted MRI after gadolinium contrast medium injection showed strongly enhanced neomembranes (Fig. 2b). We diagnosed him with a subarachnoid and intraparenchymal abscess that was mainly localized in left insula cortex and the Sylvian fissure. Additionally, contrast-enhanced chest CT demonstrated multiple small nodular shadows at the bilateral lung parenchyma, suggesting a lung abscess.

We performed surgical excision and drainage to avoid a worsening infection. The intraoperative view revealed that the Sylvian fissure was filled with purulent matter that was spreading on the surface of the brain (Fig. 2c). The intraparenchymal pus collection in the left insula cortex also communicated with that in the Sylvian fissure. Arteries running in the Sylvian fissure showed discoloration to milky white and partial redness. Just after the surgery, the patient began heavy pulsatile epistaxis from the right side. We had difficulty stopping the bleeding, which consequently required intranasal gauze packing.

Although he recovered and became alert with a slight motor weakness on the right side, severe left hemiparesis and semicoma appeared suddenly on day 8. A massive ICH occurred in the left frontotemporal lobe (Fig. 2d). A head CT angiogram (CTA) showed that fusiform aneurysms developed in all trifurcations of the left middle cerebral artery (MCA; Fig. 2e), which were approximately 9 mm × 6 mm in diameter in the dilatation of the upper branch of the M2 MCA segment. We recognized that the aneurysm had progressively enlarged and soon ruptured as a result of the subarachnoid abscess. Considering the mass effect of hemorrhage, we performed trapping and resection of the ruptured branch between the proximal M2 and M3 branches to prevent aneurysmal re-rupture. Histopathological examinations showed disruption of the intima-media structure and dissection in the media resulting from inflammatory cell infiltration, abscess, and necrotic matter (Fig. 2f). Postoperative MRI showed cerebral infarction in the regional left frontal and temporal lobes. A head angiogram performed 6 months later showed degeneration of the left MCA.

Additionally, follow-up MRA and CTA images revealed that an irregular aneurysmal mass that projected into the right sphenoidal sinus had become progressively enlarged. We considered that the heavy epistaxis after the first surgery had resulted from a rupture of the right cavernous carotid aneurysm, and a sphenoid sinus infection had caused the progressive enlargement of the aneurysm.

A stent-assisted coil embolization was attempted under general anesthesia on day 20. Plasgrel (20 mg) was administered just before the procedure. A control three-dimensional rotated angiogram revealed irregular-shaped aneurysms in the cavernous segment of the right ICA with complex components (Fig. 3a, b, and c) in the medial dome, which had a maximum diameter of 10 mm, and the lateral dome, which had a maximum diameter of 5 mm. A LVIS™ stent (3.5 mm × 22 mm; MicroVention-Terumo, Tustin, CA, USA) was overlapped into the Neuroform Atlas™ stent (4 mm × 21 mm: Stryker Neurovascular, Fremont, CA, USA) with the expectation of flow obliteration to reduce blood flow to the aneurysm (Fig. 3f). Postoperative angiography (Fig. 3d, e) showed effective packing. Aspirin (100 mg) and clopidogrel (75 mg) were administered daily starting the day after the procedure. DW-MRI revealed only a few asymptomatic new ischemic spots. The post-operative course was uneventful, and a follow-up angiography showed no recurrence of the aneurysm 6 months later.

On admission, meropenem and linezolid were administered.