



Case Reports

Mycotic intracranial aneurysms rupture presenting as pure acute subdural hematoma in infectious endocarditis. Report of 2 cases and review of the literature



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ABSTRACT

Acute subdural hematoma (ASDH) revealing mycotic aneurysm (MA) is an exceptional occurrence.

We report 2 cases of MA-related pure ASDH in the course of infective endocarditis (IE) without history of head trauma, hypertension or coagulopathy.

Case 1: A 54-year-old man presented with a 10-day history of headache, fever. At admission neurologic examination and Brain-CT were normal. Blood cultures showed *Streptococcus bovis*. MRI 5 days later revealed ischemic spots and minime ASDH over the right convexity. Cerebral angiography, revealed a 3 mm saccular aneurysm at the bifurcation of a distal branch of the right posterior cerebral artery (PCA).

Case 2: A 42-year-old man presented with a 8-day history of fever, and worsening headache. Mitral regurgitation was evidenced. A *methicillin-sensitive staphylococcus aureus* left-sided IE was diagnosed. Pre- and post-contrast Brain-CT were normal. He presented 15 days later a secondarily generalized status epilepticus. Brain CT showed an ASDH over the convexity with falco-tentorial extension, midline shift and temporal herniation. It was removed in emergency. Cerebral angiography revealed a MA of a peripheral branch of the left PCA.

The 2 patients were successfully treated by endovascular glue embolization and recovered without complications.

Even rare, ruptured MA should be considered as a cause of pure ASDH of unknown origin.

As MA can be missed on CT- and MR-Angiography because of mass effect, cerebral angiography may be mandatory.

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1. Introduction

Mycotic aneurysms (MAs) account for <5% of intracranial aneurysms and 65% of patients with MAs have underlying endocarditis [1,2]. Manifestation of a ruptured MA with acute subdural hematoma (ASDH) is an extremely rare occurrence in the course of IE and to date only 8 cases have been reported (Table 1) [3–10].

From a prospectively collected database of patients who were admitted with IE to our Institution from January 2006 to January 2017 (297 patients), 15 patients had MAs (5%), two of them presenting with ASDH without associated SAH, also named pure ASDH are reported here (13.3%).

2. Cases

2.1. Case report 1

A 54-year-old man with a 10-day history of headache, fever and diffuse joint pains was admitted at our Institution. His physical examination revealed a II/VI systolic heart murmur and a II/VI Aortic insufficiency murmur. Non-contrast Brain CT at admission was normal. The patient was started on broad-spectrum antibiotic therapy.

Blood examination showed a white blood cell count of 14000/mm³ (80% PNN). The results of other laboratory tests were within normal range limits. Blood cultures were positive for *Streptococcus bovis*. An antibiotic treatment by Amoxicillin (12 g/d) and gentamycin (210 mg/d) was initiated.

Transthoracic echocardiography (TTE) showed a severe aortic regurgitation, left ventricle dilatation and 3 vegetations of 12 mm in diameter on the aortic valve leaflet (tricuspid valve). Brain MRI

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Table 1

Cases of Acute Subdural Hematoma (ASDH) due to Mycotic Aneurysm (MA) rupture in Infective Endocarditis (IE).

	Author (year) (Reference)	Age/ Sex	Valve	germ	MA Location	SAH	ICH	Treatment	Outcome
IE/AM + SDH alone									
1	King (1960) [3]	23/F	MVN	Streptococcus pyogenes	Distal MCA			Evacuation, resection	Good recovery
2	Kasuya (1985) [4]	13/F	MVN	NA	Distal PCA			Evacuation, conservative	Good recovery
3	Present Case 1 (2018)		AVN	Streptococcus bovis	Distal PCA				
4	Present Case 2 (2018)		MVN/ AVN	Staphylococcus aureus	Distal PCA				
IE/AM + SDH + SAH									
5	Barami (1994) [7]	36/M	MVN/ AVN	NA	Distal PCA	1		Evacuation, conservative	Vegetative state
6	Matsuda (2002) [9]	22/F	MVN	NA	Distal PCA	1		Coil embolization	Good recovery
IE/AM + SDH + ICH									
7	Bamford (1986) [5]	50/M	MVN	Streptococcus viridans	Distal MCA		P,O	Evacuation, resection	Good recovery
8	Yamakawa (2003) [10]	21/F	MVN	NA	Distal MCA		F, P	Evacuation, resection	Vegetative state
IE/AM + SDH + SAH + ICH									
9	Bandoh (1987) [6]	26/F	NA	Streptococcus fecalis	Distal MCA	1	P,O	Evacuation, resection	Died 5 days
10	Sugino (2002) [9]	65/F	MVN	NA	Distal MCA	1	P,O	Conservative	Died
IE + SDH without MA									
11	Yanagihara (2003) [16]	54/M	MVN	Staphylococcus aureus	Multiple Abscesses			Evacuation	Hemiplegic
12	Rasmussen (2014) [17]	81/M	MVN- AVP	Lactococcus Garvicae				Evacuation	Good recovery
13	Geissenberger (2015) [18]	38/M	AVN	Streptococcus Mitis- oxytoca			P	Evacuation	Died

SAH: Subarachnoid hemorrhage; ICH: Intracerebral hematoma; M: Male; F: Female; MVN: Mitral valve native; MCA: Middle cerebral artery; NA: not available; PCA: Posterior cerebral artery; AVN: Aortic valve native; P: Parietal; O: Occipital; F: Frontal; AVP: Aortic valve prosthetic.

showed a minime right-sided subdural hematoma without subarachnoid hemorrhage (SAH), intracerebral hematoma or microbleeds and 3 ischemic spots (2 in the left frontal and parietal cortex, 1 in the left semi-ovale center) (Fig. 1-a-b). Post-contrast volumic 3D T1 MRI sequence and magnetic resonance angiography (MRA) were unremarkable. Catheter angiography 4 days later revealed a 3 mm saccular aneurysm at the bifurcation of a distal branch of the right posterior cerebral artery (PCA) which was treated by endovascular procedure (Fig. 1-c-d). Control MRI/post-contrast MRA 1 and 2 months later did not evidence recurrent or new MA, parenchymal sequelae or asymptomatic lesion

Thoracic and abdominal CT-angiography (CTA) revealed splenic and cardiac emboli. Cardiac-MRI showed an acute anterior infarct of the left ventricle. Furthermore, 3 extracranial MAs (superior gluteal and popliteal arteries) were successfully treated.

During the same period, the patient underwent aortic valve replacement.

3. Case report 2

A 42-year-old man, with a history of intravenous drug abuse, and of *methicillin-sensitive staphylococcus aureus* (MSSA) IE one year before was admitted at a peripheral hospital for fever, nausea and vomiting. Physical examination was normal and TTE revealed a minime mitral regurgitation without vegetation. He was admitted in our Institution 8 days later with worsening headache. Physical examination revealed petechial bleeding on the trunk, face, and subungual hemorrhages, suggesting a diagnosis of IE. Neurological examination was unremarkable. TTE and transesophageal echocardiography (TEE) revealed an increase of the mitral regurgitation, and vegetations of the tricuspid aortic valves.

He was started on broad-spectrum antibiotics (Oxacillin, 2g × 6/d, Gentamycin 210 mg/d). Blood cultures were positive for

MSSA. Pre- and post-contrast Brain CT were normal. The following day he presented an episode of partial seizures with secondarily generalized status epilepticus.

Non-contrast CT revealed a left-sided 12 mm ASDH over the left convexity, extending both to the cerebri and cerebellar falx (Fig. 1-e-f). The patient underwent urgent craniotomy and the ASDH was removed.

The day after, catheter angiography showed a fusiform aneurysm of a distal branch of the left PCA which was successfully treated (Fig. 1-g). After 4-week antibiotic therapy, MRI revealed a minime convexal and falcotentorial SDH (Fig. 1-h-i).

Common findings: The 2 patients had no history of head trauma nor hypertension or coagulopathy. MAs were successfully treated by endovascular glue embolization with good outcome. They completely resolved on immediate post-embolisation control angiography. Clinical and MRI follow-up at 1-year and 6-year for case 1 and 2 respectively, did not evidence recurrence, parenchymal sequelae or asymptomatic lesion.

4. Discussion

The main cause of spontaneous ASDH, with or without associated SAH is aneurysmal rupture with an incidence ranging between 1.8% and 3.7% in recent studies [11,12].

In our prospective cohort of 15 MAs from which these 2 patients belong, the prevalence of MA-related ASDH was of 13.3%, whereas the prevalence of MA in our series is the same as reported elsewhere [1,2].

This prevalence is greater than the prevalence of ASDH related to non-mycotic aneurysmal rupture, as detailed previously. The discrepancy between the patients included in each report is an issue to fully determine this prevalence.

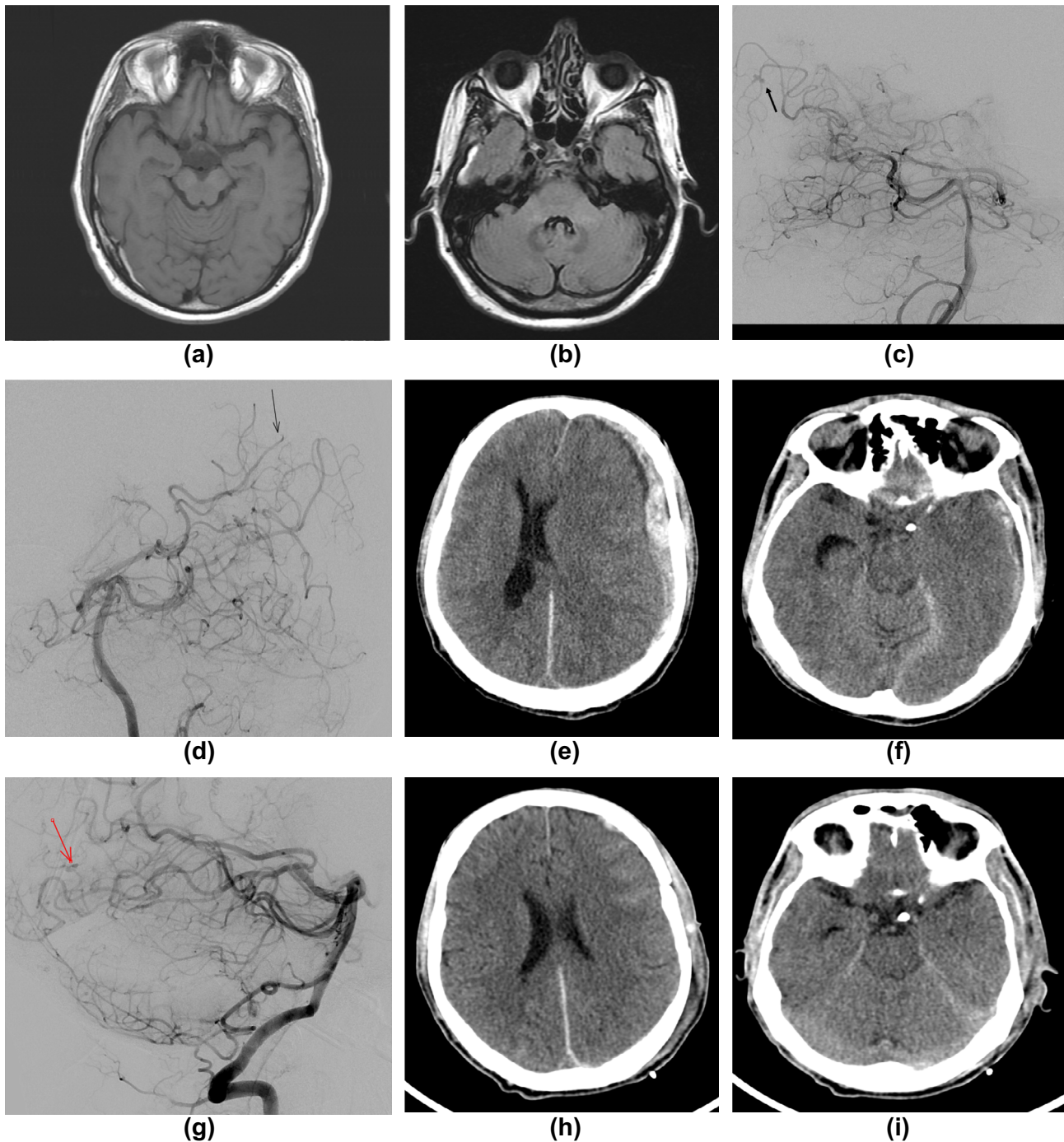


Fig. 1. Case 1: a-b-c-d: MRI shows on T1 (a) and on FLAIR sequence (b) a minime acute subdural hematoma over the right convexity without midline shift and without associated SAH. Digital subtraction angiography (DSA) reveals a sacciform mycotic aneurysm of a distal branch of the right PCA (c, arrow). Control DSA after endovascular treatment (glue embolization) shows the exclusion of the mycotic aneurysm (d). **Case 2: e-f-g-h-i:** -Non-contrast Brain-CT shows a left-sided 12 mm acute subdural hematoma at the left convexity extending along the cerebral falx and the left tentorium with 17 mm midline shift and concurrent subfalcine and transtentorial herniation. There is no evidence of brain or sub-arachnoid hemorrhage (e-f). Digital subtraction angiography (DSA) shows a fusiform mycotic aneurysm of a distal branch of the left PCA (g). Follow-up CT shows the resolution of the ASDH (h-i).

MAs are relatively rare complications of IE, found in only 2% to 4% of patients [1]. They occur more frequently in the course of acute IE, *Streptococcus viridans* and *Staphylococcus aureus* being the most frequently associated microorganisms [13]. MAs have characteristic radiological features. They develop rather at a peripheral branch than at a proximal one. They are located in the MCA (57.4%) and in the CPA (17.6%) or their distal branches. Mortality is as high as 12–32% in the recent literature reviews [2]. Furthermore, 92.7% of patients presenting with MA had a vegetation,

with the mitral valve being the most frequent location [14]. Headache is the most frequent onset symptom and mitral regurgitation the most frequent associated valve lesion.

Pure ASDH due to aneurysm rupture is extremely rare with only 45 cases published since 1981 [15]. Our literature review of IE-related MA and a concurrent ASDH shows that only 11 cases, including ours, have been reported to date (Table 1). The paucity of MA-related ASDH cases of the literature and the limited data reported impairs a rigorous comparison.

ASDHs revealing MA occur mainly in young patients with a range 2 months – 65 years (mean: 32). Only 4 pure ASDH have been reported in this context (36.4%) (3–4, reported cases). ASDH was associated with SAH (1 case, 8), CH (4 cases) [5,6,7,9] and with both SAH and cerebral hemorrhage (2 cases) [6,9]. In 8 cases where valve lesion was specified the mitral valve was concerned in 7, the aortic valve in one case, and association of both in 1 case. The distal branches of the MCA were more frequently involved (54.5%) than PCA ones (45.5%). In only 5 cases the microorganism is reported: *Streptococcus* in 4 (pyogene, viridans, fecalis, bovis) and MSSA in a last case.

Interestingly, ASDH occurring in the course of acute IE without associated MA have been reported in 3 well documented cases (Table 1) [16–18]. In Yanagihara's case [16] an ASDH over the convexity, extending to the interhemispheric cistern was associated with extended SAH. Catheter angiography did not detect any aneurysm and 20 days later multiple abscesses were observed. In Rasmussen's case [17] *Lactococcus garvieae* was identified in blood cultures in a 2-month-old infant, and vegetations were evidenced on native mitral valve and on prosthetic aortic valve. The operating findings showed a large temporal hematoma in continuity with the ASDH. In Geissenberger's autopsied case [18], where *Streptococci* of the mitis group and *Klebsiella oxytoca* were isolated from the aortic valve, an ASDH originated from a subcortical hemorrhage which had perforated into the subdural space. However, arterial rupture may result from necrotizing arteritis consecutive to septic embolism, without the presence of an aneurysm.

Since the peripheral MAs may be missed on CTA/MRA because of mass effect, a cerebral angiography is recommended.

Many mechanisms have been proposed to explain the ASDH formation in case of aneurysmal rupture. Our 2 cases, as well as other reports in the literature [7,10,18], suggest that in case of MA two of the evoked mechanisms may explain ASDH formation: If the ruptured vessel adheres to the arachnoid membrane, or if the blood streaming out of the leak disrupts the arachnoid forcing its way into the subdural space. These conditions are present in case of MA of M4 (MCA) segment and P4 (PCA) segment, as in our patients, close to arachnoid surface may favor ASDH.

This mechanism can explain an increase prevalence of ASDH in MA context.

5. Conclusion

Even rare, ruptured MA should be considered as a cause of pure ASDH of unknown origin. The necessity to repeat CTA/MRA to detect intracranial MAs in IE associated ASDH is emphasized. As MA might have been missed on CTA and MRA because of mass effect, cerebral angiography may be mandatory.

Conflict of interest

The authors have no conflict of interest and have not received any financial support. Informed consent was obtained from the 2 patients.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.jocn.2018.12.035>.

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