Our experience in the diagnosis and treatment of cerebral pseudoaneurysms

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Received 12 October 2010; accepted 5 January 2011

KEYWORDS
Pseudoaneurysm; Endovascular; Neuroradiology; Mycotic; Iatrogenic; Traumatic

Abstract
Objective: To present our experience in the diagnosis and intravascular treatment of cerebral pseudoaneurysms.

Material and methods: We present 11 pseudoaneurysms (2 traumatic, 2 mycotic, 3 iatrogenic, and 4 with other causes). We analyze the methods and diagnostic criteria, radiological and clinical outcome, the criteria used in making decisions about treatment, the method of treatment, and the complications.

Results: Digital subtraction angiography is the gold standard for the diagnosis of cerebral pseudoaneurysms; the diagnostic criteria in the literature include: aneurysms with early morphological changes and distal aneurysms or proximal aneurysms associated with another distal one, in the context of the right symptoms and signs. In the nine patients treated with endovascular techniques, the treatment objective was achieved and rebleeding did not occur.

Conclusions: In cases with clinical suspicion of a pseudoaneurysm, the patient should undergo angiography. This is especially important in patients with inexplicable cerebral hemorrhage and in those with septicemia. CT angiography and MR angiography have good diagnostic accuracy and can replace conventional angiography. However, the treatment of choice is endovascular and treatment should not be delayed unless access to the pseudoaneurysm is impeded, usually due to severe cerebral vasospasm.

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Nuestra experiencia en el diagnóstico y tratamiento de los seudoaneurismas cerebrales

Resumen
Objetivo: Presentar nuestra experiencia en el diagnóstico y tratamiento intravascular de los seudoaneurismas cerebrales.

Material y métodos: Presentamos 11 casos de seudoaneurismas (2 traumáticos, 2 micóticos, 3 iatrogénicos y 4 asociados a otras causas) en otros tantos pacientes y analizamos los métodos y criterios diagnósticos, la evolución radiológica y clínica, los criterios tomados en cuenta para la decisión terapéutica, el método de tratamiento y las complicaciones.

Resultados: El método de referencia para el diagnóstico es la angioografía por sustracción digital y los criterios diagnósticos en la literatura médica incluyen: aneurismas con cambios morfológicos precoces, aneurismas distales o aneurismas proximales asociados a otro distal, en el contexto clínico adecuado. En los 9 pacientes tratados mediante técnica intravascular se consiguió el objetivo del tratamiento, evitar el resangrado.

Conclusiones: Ante la sospecha clínica de un seudoaneurisma todo paciente debe ser valorado mediante arteriografía, principalmente aquellos que presentan hemorragias cerebrales inexplicables y los pacientes con septicemia. La angioografía mediante TC o mediante RM puede sustituir a la arteriografía con una buena rentabilidad diagnóstica. El tratamiento de elección debe ser el intravascular de primera intención y no demorarse excepto que exista una razón que impida el acceso al seudoaneurisma, normalmente un vasoespasmo cerebral grave.

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Introduction

A pseudoaneurysm is a cavity of an encapsulated organized hematoma communicating with a vessel lumen that occurs when there is a rupture in the 3 layers of the wall. This classic definition includes mainly those entities with a mycotic or traumatic origin. The concept is usually extended to include any cerebral vascular dilation not matching the criteria of a true aneurysm, and would therefore include typical pseudoaneurysms, lesions associated with rupture of arteriovenous malformations, blister-like aneurysms (secondary to carotid wall thinning giving an image similar to a blister), iatrogenic aneurysms and dissecting aneurysms. The most common clinical manifestation is unexplained brain hemorrhage, secondary to a traumatic injury or found in atypical locations.1

Management of these entities is a challenge given the initial intention of preserving the damaged vessel and to the fact that, on some occasions, standard endovascular and microsurgical techniques are not possible.1-4

We present our experience in the diagnosis and endovascular management of 11 patients with cerebral pseudoaneurysms.

Material and methods

With prior approval from our hospital ethics committee, we present 11 patients with pseudoaneurysms (Table 1). We analyze the methods and diagnostic criteria, clinical manifestations, radiological outcome, criteria in therapeutic decision-making, modality of treatment (written informed consent was obtained from all patients) and complications during the procedure. We have prospectively reviewed the patient information in our database gathered during the last 5 years. Progression was assessed by angiography performed at 6, 12 and 24 months and completed through personal interviews during outpatient visits using the modified Rankin scale (mRS) at 1 year with analysis of the score. Diagnostic criteria are shown in Table 2.

Results

The first of the traumatic pseudoaneurysms presented as a delayed hemorrhage (48 h following traumatic injury) and the second one as a severe subarachnoid hemorrhage (SAH) within the setting of a traumatic brain injury. The diagnosis was obtained by computed tomography angiography (CTA) and confirmed by angiography. Early endovascular treatment was performed and complications due to cerebral vasospasm showed: in case 1 due to a thrombus in the M1 segment of the middle cerebral artery (resolved during surgery using mechanical fibrinolysis), and in case 2 the spasm prevented performing occlusion and full treatment during the first session. Long-term follow-up showed stability in the treatment and patients progressed satisfactorily (Figs. 1 and 2).

Clinical suspicion of mycotic pseudoaneurysms (cases 3 and 4) was presented in a septic patient with an unexplained brain hemorrhage. Diagnosis was confirmed by angiography since CTA was negative. Pseudoaneurysms were found in distal branches and the early endovascular treatment was aimed at branch occlusion: platinum coils were used in the temporal artery and as for the distal posterior circulation, spontaneous disappearance of the pseudoaneurysm was observed. Both patients responded favorably (Figs. 3 and 4).

In cases 5 and 6 the clinical suspicion of a blister-like aneurysm in one case and a distal flow-related aneurysm of the basilar artery bifurcation in the other one (since there was bilateral carotid stenosis) existed. Associated with
<table>
<thead>
<tr>
<th>Number</th>
<th>Sex/age</th>
<th>Previous history</th>
<th>Diagnosis</th>
<th>Etiology</th>
<th>Hemorrhage</th>
<th>Location</th>
<th>Treatment</th>
<th>Prognosis (mRS within a year)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Male/22</td>
<td>No</td>
<td>CTA</td>
<td>Traumatic</td>
<td>Hunt–Hess IV SAH</td>
<td>Supraclinoid</td>
<td>Endovascular remodeling</td>
<td>mRS 1</td>
</tr>
<tr>
<td>2</td>
<td>Male/42</td>
<td>No</td>
<td>CTA</td>
<td>Traumatic</td>
<td>Delayed intra-</td>
<td>ACA</td>
<td>Endovascular remodeling</td>
<td>mRS 1</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>parenchymatous</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>Female/37</td>
<td>No</td>
<td>Angiography</td>
<td>Mycotic</td>
<td>Sylvian hematoma</td>
<td>Temporal anterior artery</td>
<td>Endovascular coiling</td>
<td>mRS 1</td>
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<tr>
<td>4</td>
<td>Male/50</td>
<td>No</td>
<td>CTA normal positive angiography</td>
<td>Mycotic</td>
<td>Intraventricular hemorrhage</td>
<td></td>
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<td>mRS 0</td>
</tr>
<tr>
<td>5</td>
<td>Male/47</td>
<td>No</td>
<td>Angiography</td>
<td>Associated to blister type</td>
<td>Hunt–Hess III SAH</td>
<td>Supraclinoid</td>
<td>Endovascular remodeling (stent)</td>
<td>mRS 1</td>
</tr>
<tr>
<td>6</td>
<td>Female/65</td>
<td>HBP, diabetes</td>
<td>Angiography</td>
<td>Associated to true aneurysm</td>
<td>Hunt–Hess III SAH</td>
<td>Top of the basilar</td>
<td>Endovascular remodeling (coiling)</td>
<td>mRS 1</td>
</tr>
<tr>
<td>7</td>
<td>Male/42</td>
<td>HIV+, HCV+, IVDU</td>
<td>CTA</td>
<td>AVM associated to nidal rupture</td>
<td>Perimesencephalic SAH and posterior fossa</td>
<td>Posterior fossa AVM</td>
<td>Endovascular remodeling (Onyx)</td>
<td>mRS 0</td>
</tr>
<tr>
<td>8</td>
<td>Male/42</td>
<td>Former IVDU, smoker</td>
<td>CTA</td>
<td>AVM associated to a flow-related aneurysm</td>
<td>Cerebral intra-parenchymatous hemorrhage</td>
<td>Posterior fossa AVM</td>
<td>Endovascular remodeling (Onyx)</td>
<td>Death</td>
</tr>
<tr>
<td>9</td>
<td>Male/45</td>
<td>Hypophysis macroadeno, panhypopituitarism</td>
<td>CTA</td>
<td>Iatrogenic</td>
<td>Hunt–Hess III SAH</td>
<td>Top of the basilar</td>
<td>No</td>
<td>Death</td>
</tr>
<tr>
<td>10</td>
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<td>Angiography</td>
<td>Iatrogenic</td>
<td>SAH Hunt–Hess III</td>
<td>Supraclinoid</td>
<td>Endovascular remodeling</td>
<td>Death</td>
</tr>
<tr>
<td>11</td>
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<td>Angiography</td>
<td>Iatrogenic</td>
<td>SAH Hunt–Hess II</td>
<td>Left A1–A2</td>
<td>Endovascular remodeling</td>
<td>mRS 2</td>
</tr>
</tbody>
</table>

A1–A2: union of segment A1 and A2 of the ACA; ACA: anterior cerebral artery; IVDU: intravenous drug user; CTA: computed tomographic angiography; SAH: subarachnoid hemorrhage; HBP: high blood pressure; Hunt and Hess: Hunt and Hess scale for SAH grading by CT; mRS: modified Rankin scale for prognostic grading; Onyx: liquid embolic agent; remodeling: balloon assisted embolization; TOP of the basilar: aneurysm located at the distal bifurcation of the basilar artery; HCV: positive serology for hepatitis C virus; HIV+: positive serology for human immunodeficiency virus.
Table 2 Arteriography diagnostic criteria for the diagnosis of cerebral pseudoaneurysms.

<table>
<thead>
<tr>
<th>Diagnostic criteria for cerebral aneurysms: angiography</th>
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<tbody>
<tr>
<td>Morphologic</td>
</tr>
<tr>
<td>1. Contrast agent collection adjacent to a cerebral vessel, showing contrast retention and delayed washout²</td>
</tr>
<tr>
<td>2. Occlusion or stenosis adjacent to the aneurysm in the adequate clinical setting</td>
</tr>
<tr>
<td>Temporal Location</td>
</tr>
<tr>
<td>Cerebral aneurysm presenting significant morphologic changes in a short period of time²,³</td>
</tr>
<tr>
<td>Etiologic</td>
</tr>
<tr>
<td>Traumatic aneurysms in patients with open traumatic injuries. In closed injuries if unexplained or delayed cerebral hemorrhage exists or lethargy, coma, massive epistaxis, paralysis of cranial pairs or cranial base fractures an angiography or CTA will be performed</td>
</tr>
<tr>
<td>Mycotic: endocarditis or septicemia with positive blood culture associated to the presence of an aneurysm in a distal branch (segments 2, 3 or 4 of the middle or posterior cerebral artery) or in a proximal branch with the morphologic changes described</td>
</tr>
</tbody>
</table>

Figure 1 Case 1: 22-year-old male with traumatic pseudoaneurysm diagnosed by the presence of a massive SAH. (A) Unenhanced cranial CT: traumatic brain injury with facial fractures associated with extensive SAH due to a cerebral pseudoaneurysm. (B) Cerebral angiogram: supraclinoid pseudoaneurysm with vasospasm at the branches of the MCA. (C) Cerebral angiogram: following occlusion of the pseudoaneurysm using platinum coils (coiling).

These findings, the angiography showed accumulation of contrast with the morphological variations typical of pseudoaneurysms. Both cases were treated with endovascular techniques: in the blister-like type by delaying the treatment to the subacute phase in order to be able to place a stent, and in the case of the posterior circulation aneurysm through coil embolization with the objective to treat only the proximal aneurysm, to avoid intraoperative ruptures.

The rupture of an arteriovenous malformation (AVM) can result from complications related to the venous drainage

Figure 2 Case 2: 42-year-old male with delayed hemorrhage following traumatic brain injury. (A) Cerebral angiogram: posttraumatic pseudoaneurysm of the anterior cerebral artery (ACA) diagnosed by delayed intraparenchymatous hemorrhage. (B) Cerebral angiogram: contrast retention and delayed washout image typical of cerebral pseudoaneurysms. (C) Cerebral angiogram: endovascular remodeling with coiling (intra-arterial balloon assisted).
or the presence of associated aneurysms. In two of the cases shown, aneurysms typical of this entity diagnosed by angiography were present (a flow-related aneurysm and a nidal aneurysm) but showing the characteristic features of pseudoaneurysms (contrast retention, delayed washout and morphological changes). Since the pedicle facilitated the reflux in these two cases, they were treated in the acute phase with Onyx (a liquid embolic agent used for endovascular occlusion of certain conditions such as AVMs that requires a security distance to the catheter tip since before advancing it occludes the pedicle proximally) with good angiographic results.

This series also includes 3 cases of iatrogenic pseudoaneurysms. One of them occurred during transsphenoidal surgery of a macroadenoma that damaged an aneurysm at the distal bifurcation of the basilar artery, diagnosed by CTA and angiography. Early endovascular treatment was considered, but could not be performed since the first angiography showed spontaneous thrombosis and the patient died from postoperative complications. Regarding the two cases of intraoperative ruptures during endovascular treatment, diagnosis was achieved at the follow-up angiography performed 7 days after the rupture and in both cases were treated with endovascular techniques using balloon remodeling and embolization with platinum coils in the acute phase with good angiographic results (Fig. 5). One of the patients died from the initial hemorrhage and the other recovered satisfactorily.

Discussion

Traumatic pseudoaneurysms are a rare entity that represent less than 1% of all cerebral aneurysms with mortality rates as high as 50%. In up to 40% of the cases they occur following an open traumatic injury or a closed one in 3% secondary to vessels injured by the bone or dura mater. It has been more frequently described in pediatric population. Early diagnosis and treatment are necessary. Digital angiography increases the sensitivity and specificity of CTA. If the initial test is negative it is advisable to repeat it from after two weeks up to a month. Series of surgical treatment with a morbidity between 18% and 29% and small series of endovascular management without

Figure 3  Case 3: 3-year-old female with sepsis of respiratory origin. (A) Unenhanced cranial CT: spontaneous sylvian hematoma. (B) Cerebral angiogram: pseudoaneurysm of the anterior temporal artery, causing sylvian bleeding. (C) Cerebral angiogram: endovascular treatment with occlusion using platinum coils of the aneurismatic sac and branch occlusion.

Figure 4  Case 4: 50-year-old male with respiratory sepsis and spontaneous hemorrhage. (A) Unenhanced cranial CT: intraventricular hemorrhage. (B) Cerebral angiogram: pseudoaneurysm of the left parieto-occipital artery with SAH and respiratory sepsis. (C) Cerebral angiogram: spontaneous resolution within 15 days following antibiotic therapy.
rebleeding or mortality with less than 10% of morbidity also have been described; thus endovascular management seems to improve the natural history of this disease. In our series, the clinical–radiological suspicion was essential, and the presentation was typical. Early endovascular treatment was performed with excellent results despite the associated cerebral vasospasm.

Infections are the second most common cause. Mycotic pseudoaneurysms are caused by necrosis and rupture of the artery wall secondary to septic emboli. Series with an incidence of pseudoaneurysms between 1% and 10% in all endocarditis have been described. In these cases, it is necessary to perform a screening angiography, that might be replaced by a CTA, and repeat it only in case of neurological symptoms. Behavior of these lesions is unpredictable, and growth, regression, disappearance or rupture of the aneurysm is seen at follow-up. Predictive signs of aneurysmatic rupture of questionable usefulness have been identified related to this matter. Mortality in mycotic pseudoaneurysms ranges between 20% and 83% in spite of antibiotic treatment. Mortality in the series of patients treated with surgery of the hematoma and antibiotics ranges between 7% and 61%. In series assessing the endovascular treatment, mortality values are close to 0% with a morbidity of 35% related to the initial ictus. Within a septic setting, patients 3 and 4 presented cerebral hemorrhages of undetermined etiology diagnosed as distal mycotic pseudoaneurysm, with the approach of performing endovascular treatment for vascular occlusion, although one of them recovered spontaneously.

Other more rare causes of cerebral pseudoaneurysms are iatrogenic aneurysms, those associated with drug consumption, associated with true aneurysms, secondary to AVM nidus rupture, oncotic aneurysms secondary to tumor embolization and, lastly, spontaneous or idiopathic pseudoaneurysms. Lesions that match the criteria for pseudoaneurysms associated to a true aneurysm must follow early treatment of the aneurysm avoiding flow of material to the pseudoaneurysm to prevent intraoperative ruptures. The diagnosis of pseudoaneurysms associated with AVM ruptures (cases 7 and 8) requires a suspicious CTA and an angiography meeting the diagnostic criteria for a pseudoaneurysm. If these findings are confirmed, early treatment must be followed and should not be delayed depending on the progression of the hemorrhage. In our experience, when facing an intraoperative rupture during endovascular surgery, a control angiography should be performed within a week, even without evidence of rebleeding, since the presence of an associated pseudoaneurysm might be diagnosed thus worsening the prognosis of the patient. In the event of surgery, an early CTA should be performed.
Our experience in the diagnosis and treatment of cerebral pseudoaneurysms

Regarding the global treatment of these entities, the main objective is to avoid rebleeding. The few published series claim that endovascular treatment seems to improve results concerning the catastrophic natural history of this condition.1-4 In general terms, these series claim that early treatment is fundamental to improve the natural history of this condition. In pseudoaneurysms associated with cerebral vasospasm, the series agree that early treatment must be delayed until the vasospasm has resolved.1-4 Regarding what technique to follow, the main factor under consideration must be the location of the pseudoaneurysm: carotid ones and those located in segment 1 of the middle, anterior and posterior cerebral artery are proximal aneurysms. Treatment is the same as in normal saccular aneurysms by using coils or stents, even in the acute phase. If it is not possible to close the aneurysm in a proximal branch, an occlusion test should be performed prior to closing the branch and it is advised to always perform it in conspicuous areas even if it is in distal branches. At present, Onyx is a very useful and manageable tool, mainly in those lesions associated with an AVM and with sufficient segment for Onyx reflux.1,4,6-13,17 Distal aneurysms are treated with vascular occlusion that includes the aneurismal sac, if possible. Coils, glue or embolic agents are used to this end.1,4,6-10

Regarding prognosis, in the 9 patients treated with endovascular techniques, the treatment objective was achieved and rebleeding did not occur. Follow-up showed good progression with treatment stability in the 8 patients who survived. In the only patient who underwent parent artery occlusion (case 3), the imaging tests did not show ischemic lesions, although they were probably hidden by a sylvian hematoma and postoperative changes. The patient showed minor hemiparesis as a sequel. No signs of rebleeding were found in the 3 patients who died; patient 9 died from medical complications from an underlying endocrinological disease that worsened by hemorrhage, patient number 8 died in the setting of an acute hydrocephalus and number 10 due to the intraparenchymatous hematoma and increased intracranial pressure. The only patient (case 4) showing spontaneous favorable progression in a pseudoaneurysm of distal posterior circulation and resolution of bacteremia with antibiotics showed absence of recanalization at long term follow-up.

In conclusion, pseudoaneurysms are rare pathological entities associated to high morbidity and mortality that require an early clinical and/or radiological diagnosis. In cases with clinical suspicion of a pseudoaneurysm, the patient should undergo angiography, although CTA or magnetic resonance angiogram (MRA) can be useful options particularly in those patients with unexplained brain hemmorhages and in patients with septicemia. Endovascular treatment must be the primary treatment and should not be delayed unless access to the pseudoaneurysm is not possible, usually due to severe cerebral vasospasm. Moreover, interdisciplinary medical management is essential since these patients usually present multiple associated pathologies.

Authorship

1. Responsible for the integrity of the study: EMQ.
2. Conception of the study: EMQ, AGG.
3. Design: EMQ, AGG.
4. Acquisition of data: EMQ, AGG.
5. Analysis and interpretation of data: EMQ, AGG, AMM, PVV, MB, JCG, AL.
6. Bibliographic search: EMQ.
7. Drafting of the manuscript: EMQ, AMM.
8. Critical review with intellectually relevant contributions: EMQ, AGG, AMM, PVV, MB, JCG, AL.
9. Approval of the final version: EMQ, AGG, AMM, PVV, MB, JCG, AL.

Conflict of interest

The authors declare not having any conflict of interest.

References


