FATAL HEMORRHAGIC COMPLICATION FOLLOWING ENDOVASCULAR TREATMENT OF A CEREBRAL ARTERIOVENOUS MALFORMATION

Case Report and Review of the Literature

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Summary

Evaluation of the natural history of brain Arteriovenous Malformations (AVMs) including its morbidity and mortality is a crucial point in the management of patients having a cerebral AVM. The risks associated with the AVM natural history, especially regarding the occurrence of an hemorrhage, have to be compared to the risks due to the therapeutic approach. In the literature, the risk of annual bleeding of an AVM is estimated from 2 to 4%. Morbidity from AVM rupture is estimated from 13% to 50% with a risk of mortality reported from 3 to 30%. Endovascular treatment is an efficient tool in the therapy of these lesions. However, AVM embolization remains a difficult procedure. Complications of the endovascular treatment must be evaluated in relation to the potential risk associated to the AVM natural history. After AVM endovascular treatment, morbidity with permanent neurological deficit is reported in 0.4% to 12.5% of patients and mortality in 0.4% to 7.5%. In more recent reports, after brain AVM embolization, a permanent neurological deficit is estimated to occur in 9% of patients and death in 2%. Hemorrhage appears the most frequent and serious complication in the endovascular treatment of a brain AVM. We report a case of fatal hemorrhagic complication following endovascular treatment of a cerebral AVM in a 20 year old patient. This case contributes to remind that embolization, even in specialized centers with experience in the management of this pathology, can be followed by a poor and even fatal outcome. In most cases, the treatment is performed in order to protect the patient of a potential risk. Consequently, the complication of the embolization must always be carefully considered and discussed between the medical team, the patient and its family for planning the AVM endovascular treatment.

Key words: arteriovenous malformation, brain, intracranial, endovascular treatment, complication, hemorrhage.

Résumé

Complication hémorragique fatale à la suite du traitement endovasculaire d’une malformation artério-veineuse cérébrale : à propos d’un cas et revue de la littérature

L’évaluation de l’histoire naturelle et de la morbi-mortalité spontanée des malformations artério-veineuses (MAVs) cérébrales est un aspect crucial de la prise en charge des patients porteurs de ce type de lésion. Les risques associés à l’histoire naturelle d’une MAV, en particulier ceux liés au risque de saignement, doivent être comparés aux risques liés au traitement. Les risques annuels de saignement d’une MAV rapportés dans la littérature sont entre 2 et 4 %. La morbidité secondaire à la rupture de la MAV est estimée entre 13 % et 50 % avec un risque de mortalité de 3 à 30 %. Le traitement endovasculaire apparaît comme une technique efficace dans le traitement de ces lésions. L’embolisation des MAV s’est cependant une procédure difficile. Les complications du traitement endovasculaires doivent être comparées aux risques potentiels associé à l’histoire naturelle. La morbidité rapportée après traitement endovasculaire est de 0,4 % à 12,5 % des patients et la mortalité de 0,4 % à 7,5 %. Des publications plus récentes rapportent un déficit neurologique permanent après embolisation chez 9 % des patients et un décès dans 2 % des cas. Au cours du traitement endovasculaire d’une MAV cérébrale, l’hémorragie apparaît la complication la plus fréquente et la plus sèvère. Nous rapportons un cas de complication hémorragique fatale à la suite du traitement endovasculaire d’une MAV cérébrale chez un patient de 20 ans. Ce cas contribue à rappeler que l’embolisation des MAV, même au sein d’une équipe spécialisée entraînée à la prise en charge de ce type de pathologie, peut être à l’origine de complications sévères voire fatales. Dans la plupart des cas le traitement est réalisé pour protéger le patient d’un risque potentiel et les complications éventuelles liées à l’embolisation de la MAV doivent donc être soigneusement considérées, discutées au sein de l’équipe médicale et avec le patient et sa famille, avant de planifier le traitement endovasculaire.

Mots-clés : malformation artério-veineuse cérébrale, cerveau, intra-crânien, traitement endovasculaire, complication, hémorragie.

INTRODUCTION

The therapeutic management of cerebral arteriovenous malformations (AVMs) includes interventional neuroradiology, neurosurgery and radiosurgery. The risks associated with AVM therapy are justified by the poor outcome associated to the natural history of these lesions [2, 4, 17, 28, 31, 40, 45]. Thanks to improvement in endovascular technique, the endovascular approach of brain AVMs is an efficient technique. Embolization can allow a complete and definitive occlusion of the AVM or, reducing the AVM size, can permit or facilitate a
complementary treatment by radiosurgery or neurosurgery [6, 20, 33, 34, 36, 43, 47]. However, AVM embolization is still a technical challenge and remains a difficult procedure. Complications of the endovascular treatment must be evaluated in relation to the potential risk associated to the AVM natural history. We report a case of fatal complication due to endovascular treatment of a cerebral AVM in a young adult patient. In addition, we reviewed the literature concerning the AVM natural history and the complications related to AVM endovascular treatment.

CASE REPORT

A 20 year-old male patient presented with partial secondarily generalized seizures. Computed Tomography (CT) and Magnetic Resonance (MR) exams revealed a left posterior temporal ArterioVenous Malformation (AVM). Anti epileptic therapy was administered (Depakine 1000mg/day). An angiographic study performed in our department in October 2000 confirmed the diagnosis of mid-sized cerebral AVM. The lesion was located in the left inferior temporal lobe. Feeding pedicles arose from the left posterior temporal artery, branch of the posterior cerebral artery (PCA) and there was no trans-dural supply from the external carotid artery. The nidus showed an arterio-venulo-angioarchitecture and presented a vascular pouch (figure 1a). There was a single drainage into a cortical vein joining a hypoplastic left transverse sinus with a predominant retrograde drainage into the controlateral transverse sinus. A venous stenosis was observed along the course of the draining vein (figure 1a-c). On the basis of the clinical and angiographic features including the young age of the patients and the angiographic risk factors, multidisciplinary medical team concluded that treatment of this AVM was indicated and that the initial option was an endovascular treatment by embolization. After consultation with the patient and his family, the first endovascular session was planned in March 2001. The procedure was performed under general anesthesia. After positioning the guiding catheter into the left vertebral artery, embolization was performed successively through 3 pedicles using glue (N-butyl cyanoacrylate). Angiographic control of the left vertebral and internal carotid arteries showed an angiographic occlusion of the AVM (figure 1d-f). As AVMs may appear to be obliterated immediately after embolization and sometimes recanalize on follow-up, we always perform an angiographic control. The angiography, performed three months after the endovascular treatment (June 2001), showed a residual compartment of the AVM and a second endovascular session was planned in September 2001. As at first endovascular procedure, the patient was put under general anesthesia and endovascular procedure was performed using biplane angiographic equipment (Siemens, Erlangen, Germany) with high quality road map and subtracted fluoroscopy. The residual AVM was fed by pedicles of the posterior temporal artery arising from the left Middle Cerebral Artery (MCA), and there were no more direct feeders from the left PCA (figure 1g and 1h). The guiding catheter was positionned into the left internal carotid artery. Using a hydrophilic microcatheter (Flow-Rider 1.5) with a microguide, it was possible to catheterize selectively the feeding pedicle arising from the MCA (figure 1i) and thus to reach the lesion. In order to obtain the occlusion of the AVM, material used was again glue diluted with lipiodol at 25%. As usual after each glue injection, we did a resolute and fast retraction of the microcatheter. Although we only experienced a mild and not significant resistance during microcatheter retrieval, the latter broke inside the MCA as immediately showed on fluoroscopy. Control angiograms demonstrated that the distal part of the microcatheter was located in the M1 segment of the MCA extending after the bifurcation in the initial part of the inferior branch (figure 1j and 1k). This branch was occluded at its origin and a retrograde vascularisation of its tributary territory was observed through a collateral circulation via the leptomeningeal branches. Few minutes after, subsequent control angiograms showed the persistent permeability of the M1 segment of the MCA and a partial recanalization of the temporal branch (figure 1l). There were no angiographic signs suggesting a bleeding such as mass effect and/or extravasation of contrast medium. Thus the sedation was diminished for evaluation of the patient clinical condition. Neurological exam demonstrated a right hemiparesis. A MR scan (using our usual fast protocol for ischemia including diffusion-weighted images) was performed in order to evaluate the presence and extension of an eventual ischemia. MR exam showed a large hematoma in the left hemishere with mass effect and a CT scan, performed immediately after the MR exam, confirmed the hemorrhage (figure 1m-o). The patient was transferred to the neurosurgical operative room. A transient episode of anisocoria left > right regressed after 100cc of mannitol at 20%. The surgical intervention performed resulted in partial evacuation of the hematoma. An external ventricular catheter was positioned for monitoring of the Intra-Cranial Pressure (ICP). Three hours after surgery, the patient experienced an abrupt increase of the ICP associated with a left mydriasis and initial pupil dilatation on the right side. A follow-up CT scan showed increased cerebral edema with severe mass effect, but no re-bleeding. The patient underwent a second surgical procedure for emergency decompressive craniotomy, anterior temporal lobectomy and plastic enlargement of the dura mater. This second surgical intervention allowed regression of the ocular signs and normalization of the ICP. At the first day after interventions, the patient remained stable. At the second day, the increase of the ICP was controlled by mannitol and intermittent drainages. At days 3 and 4, severe oedematous reaction was responsible for episodes of abrupt and severe increase of the ICP with bilateral areactive mydriasis which were hardly controlled. Trans-cranial Doppler showed severe impairment of the cerebral circulation and diabetes insipidus developed. The patient expired five days after the embolization.

DISCUSSION

Arteriovenous Malformations (AVMs) of the brain are errors in the development of the vascula-
angiographie digitalisée, de face (hyperselectif du premier pédicule de l’artère cérébrale moyenne gauche (ACM). Deuxième session d’embolisation : angiographie digitalisée, de face (interne gauche (f)

artery show a mid-sized Arterio-Venous Malformation (AVM) located in the inferior temporal lobe; feeding pedicles arise from the left Middle Cerebral Artery (MCA). First (a-f) and second (g-o) sessions of embolization. Antero-posterior (a) and lateral (b) view angiograms of the left vertebral artery show a mid-sized Arterio-Venous Malformation (AVM) located in the inferior temporal lobe; feeding pedicles arise from the left posterior temporal artery arising from the left Middle Cerebral Artery (MCA). Second session of embolization: antero-posterior (g) and lateral (h) view angiograms of the left internal carotid artery; i hyperselective angiogram in antero-posterior view obtained during intermediate step of the catheterization of the first pedicle (c, arrow). After the first embolization session, including three glue injections, antero-posterior (d) and lateral (e) view angiograms of the left vertebral artery and lateral view angiogram (f) of the left internal carotid artery show angiographic occlusion of the AVM. Three months after the embolization, the control angiography showed a residual compartment of the AVM fed by pedicles of the posterior temporal artery arising from the left MCA. Second session of embolization: antero-posterior (g) and lateral (h) view angiograms of the left internal carotid artery; i hyperselective angiogram in antero-posterior view obtained during intermediate step of the catheterization of feeding pedicles. After injection of the glue, during retrieval of the microcatheter, the latter broke inside the MCA. Antero-posterior view angiograms of the left internal carotid artery (j) and unsubtracted images in k demonstrate the occlusion of the inferior branch of the MCA bifurcation (j, black arrow). The distal part of the microcatheter is visualized in the M1 segment of the MCA (j and k, white double arrows) and in the initial part of the MCA inferior branch (k, white arrows). A partial recanalization of the temporal branch is observed in the angiogram performed a few minutes later (l, arrow). After embolization, Computed Tomography (m, n and o) shows a large hematoma in the left hemisphere with mass effect.

First (a-f) and second (g-o) sessions of embolization. Antero-posterior (a) and lateral (b) view angiograms of the left vertebral artery show a mid-sized Arterio-Venous Malformation (AVM) located in the inferior temporal lobe; feeding pedicles arise from the left anterior temporal artery, branch of the posterior cerebral artery. A vascular pouch within the nidus is visualized (a, arrow). The AVM has a single drainage into a cortical vein joining a hypoplastic left transverse sinus with a predominant retrograde drainage into the contralateral transverse sinus. A venous stenosis is observed along the course of the draining vein and it is better visualized in a lateral hyperselective angiogram obtained during intermediate step of the catheterization of the first pedicle (c, arrow). After the first embolization session, including three glue injections, antero-posterior (d) and lateral (e) view angiograms of the left vertebral artery and lateral view angiogram (f) of the left internal carotid artery show angiographic occlusion of the AVM. Three months after the embolization, the control angiography showed a residual compartment of the AVM fed by pedicles of the posterior temporal artery arising from the left MCA. Second session of embolization: antero-posterior (g) and lateral (h) view angiograms of the left internal carotid artery; i hyperselective angiogram in antero-posterior view obtained during intermediate step of the catheterization of feeding pedicles. After injection of the glue, during retrieval of the microcatheter, the latter broke inside the MCA. Antero-posterior view angiograms of the left internal carotid artery (j) and unsubtracted images in k demonstrate the occlusion of the inferior branch of the MCA bifurcation (j, black arrow). The distal part of the microcatheter is visualized in the M1 segment of the MCA (j and k, white double arrows) and in the initial part of the MCA inferior branch (k, white arrows). A partial recanalization of the temporal branch is observed in the angiogram performed a few minutes later (l, arrow). After embolization, Computed Tomography (m, n and o) shows a large hematoma in the left hemisphere with mass effect.

Fig. 1. – Malformations Arterio-Veineuses cérébrales temporale gauche chez un patient de 20 ans.

Première (a-f) et deuxième (g-o) séances d’embolisation. L’angiographie digitalisée, de face (a) et de profil (b) de l’artère vertébrale gauche met en évidence une Malformation Artério-Veineuse (MAV) de taille moyenne située au niveau de la partie inférieure du lobe temporal gauche; les pédicules proviennent de la branche temporale inférieure du lobe temporal. On visualise une poche vasculaire au sein du nidus (a, flèche). La MAV a un drainage veineux unique vers une veine corticale qui rejoint un sinus transverse gauche hypoplasique avec un drainage rétrograde vers le sinus transverse contralateral. La veine de drainage présente une sténose veineuse qui est mieux visualisée sur une vue de profil après cathétérisme hyperselectif du premier pédicule (c, flèche). Après la première session d’embolisation qui a comporté trois injections de colle, l’angiographie digitalisée, de face (d) et de profil (e) de l’artère vertébrale gauche ainsi que la vue de profil de l’artère carotide interne gauche (j) mettent en évidence une occlusion angiographique de la MAV. Trois mois après, un contrôle angiographique met en évidence une composante résiduelle de la MAV alimentée par des pédicules de l’artère vertébrale postérieure provenant de l’artère cérébrale moyenne gauche moyenne (ACM). Deuxième session d’embolisation : angiographie digitalisée, de face (g) et de profil (h) de la carotide interne gauche et vue de face obtenue à un stade intermédiaire de la cathétèrisation du pédicule alimentant la MAV. Après injection de colle biologique, durant le retrait du micro-cathéter, celui-ci se rompt dans l’ACM. Angiographie digitalisée de face (j) et même vue de profil (k) de la carotide interne gauche mettant en évidence l’occlusion de la branche inférieure de la bifurcation de l’ACM (j, flèche noire). La portion rompue du micro-cathéter est visualisée dans le segment M1 de l’ACM (j et k, doubles flèches blanches) et au niveau de la portion initiale de la branche inférieure de l’ACM (k, flèches blanches). Une recanalisation partielle de la branche temporaire est observée sur le contrôle angiographique réalisé quelques minutes plus tard (l, flèche). Après l’embolisation, le scanner réalisé en urgence (m, n et o) met en évidence un volumineux hématome hémisphérique gauche entraînant un effet de masse.

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Natural History

In most published studies, the risk of annual bleeding of an AVM is estimated from 2 to 4% and a first hemorrhagic event is associated with an increased risk of a new bleeding. The risk of developing neurological symptoms from an AVM, usually because of hemorrhage, is cumulative increasing with patient age. In the young adult population, AVMs are significant risk factors for hemorrhagic stroke. [18, 28, 37, 40, 45].

Brown et al. [2] conducted a long-term follow-up study (mean follow-up time was 8.2 years) of 166 patients in order to define the natural history of clinically unruptured AVMs. Intracranial hemorrhage occurred in 31 patients (18%). The Authors confirmed that mean risk of hemorrhage was 2.2% per year, and the observed annual rates of hemorrhage increased over time. Mast et al. [28] reported that patients with hemorrhage at initial presentation have a higher risk of subsequent bleeding. During mean follow-up of 8.5 months for 142 patients presenting with hemorrhage and 12 months for 139 patients without hemorrhage, a new bleeding occurred in 18 (13%) hemorrhagic patients and in 3 (2%) of the no-hemorrhagic patients. The annual risk of hemorrhage was 17.8% and 2.2%, respectively. In patients presenting with hemorrhage, the risk of bleeding fell from 32.9% in the first year to 11.3% in subsequent years.

The relationship between the size of an AVM and its tendency to hemorrhage is unclear. Brown et al. [2] found that the size of the AVM was of no value in predicting AVM rupture. Spetzler et al. [38] prospectively evaluated 92 AVMs. Their results showed that small AVMs (equal or < 3 cm) presented with hemorrhage significantly more often than large AVMs (>6 cm), the incidence being 82% versus 21%. In 24 patients, intraoperative arterial pressures were recorded from the main feeding vessel(s): smaller AVMs had significantly higher feeding artery pressures than did larger AVMs, and they were associated with large hemorrhages. These Authors suggested that higher pressure in feeding vessel(s) may be responsible for higher frequency and severity of hemorrhage in small AVMs. Stefani et al. [41] reported that hemorrhage was the initial presentation in 47.6% of AVMs <3 cm, in 22.5% of AVMs ranging in size between 3 and 6 cm, and 20% in malformations >6 cm, but these differences concerning AVM size and bleeding were not statistically significant. On the contrary, Soderman et al. [37] reported that the hemorrhagic risk increases with AVM volume. Deep location of an AVM has been also reported as a significant factor associated to bleeding [37, 41]. The presence of systemic hypertension seems to be of no value in predicting AVM rupture [2]. Reported angiographic AVM findings associated with an increased risk of bleeding include existence of associated aneurysms (intramidal or flow related peduncle, proximal or distal) and some venous drainage patterns such as a deep venous drainage, single draining vein or a small number of draining veins, venous stenosis or ectasia [10, 25, 26, 40, 41, 49]. Duong et al. [10] reviewed 340 patients with cerebral AVMs from a prospective database. By statistical analysis, deep venous drainage, age and high feeding arterial/intracranial pressure were the most powerful and independent risk predictors for AVM hemorrhagic presentation. Mast et al. [28] found that deep venous drainage and male sex were also significantly associated with subsequent hemorrhages after an initial bleeding, but no significant association was found for age or AVM size. Stefani et al. [41] reported that, in their series, a single draining vein was associated with bleeding at presentation in 57.6% of AVMs, and 72.8% of the patients with venous ectasia had bleeding as initial evidence. The lesion of our patient, presenting nidal vascular ectasia, single draining vein and venous stenosis along its drainage, was considered as having angiographic risk factors for bleeding.

Morbidity associated with AVM bleeding is estimated <50% with a risk of mortality between 10 to 15% [45]. However, Brown et al. [2], among 31 patients with AVM rupture, reported death in 9 (29%) patients and significant long-term morbidity in 5 (16%); consequently, 23% of survivors (5/22 patients) had permanent morbidity. The morbidity rate for the entire series was 5.4% (9 of 166 patients with an AVM). Crawford et al. [4] studied 217 patients managed without therapy. Mean follow-up time was 10.4 years. Using life survival analyses, in untreated patients there was a 42% risk of hemorrhage, 29% risk of death, 18% risk of epilepsy and a 27% risk of having a neurological handicap seriously affecting daily activity by 20 years after diagnosis. Ondra et al. [31] have updated a series of 160 prospectively followed untreated symptomatic patients with cerebral AVMs. Mean follow-up period was 23.7 years. The mean interval between initial presentation and subsequent hemorrhage was 7.7 years. The rate of major re-bleeding was 4.0% per year, and the mortality rate was 1.0% per year. The combined rate of major morbidity and mortality was 2.7% per year. These annual rates remained essentially constant over the entire period of the study. At follow-up review of this series, 23% of patients were dead from AVM hemorrhage. There was no difference in the incidence of re-bleeding or death regardless of presentation with or without hemorrhage.

In the literature, the rupture of an AVM has been reported less severe and life-threatening than that from an intracranial aneurysm. Hartmann et al. [14] studied the morbidity of intracranial hemorrhage related to cerebral AVMs in 119 patients: 115 patients presented with hemorrhage and 27 of them suffered a second hemorrhage during follow-up; an additional 4 patients had other diagnostic symptoms but bled also during follow-up. Among the 115 patients, 54 (47%) had a favorable outcome, 43 (37%) were independent in their daily activities, 15 (13%) were moderately disabled and 3 (3%) were severely disabled or dead. None of the patients with a hemorrhage during follow-up died during the observation period. On the contrary, Hillman et al. [17] affirmed that the rupture of an AVM is as severe and devastating as that of an aneurysm. Although aneurysm rupture is more lethal, AVM rupture tends to result in more frequent neurological disability. In their series, good overall outcome was achieved respectively in 56% of patients with ruptured aneurysms and in 48% of patients with AVM and hemorrhage.
This is due to the high occurrence in AVMs of intraparenchymal hematoma which are more often responsible for neurological deficit(s) compared to subarachnoid hemorrhage. Consequently, as reported in the literature, cerebral AVM are relatively severe lesions which often have a poor or fatal outcome.

In the case of our patient, the indication for treatment was also based on the analysis of the angiographic aspect of the AVM associated to patient clinical findings including the young age. On the basis of experience and results in our multidisciplinary center, endovascular treatment appeared to be the best option for the treatment of this particular lesion. In addition, some Authors [24] have reported that seizures show a positive response to endovascular treatment in patients whose AVMs had been eliminated as well as in those who received only partial treatment. However, complications due to the endovascular treatment must be evaluated and, as stressed above, the risks associated with the natural history of an AVM have to be balanced with the risks due to the therapy.

Complications of the endovascular treatment in brain AVMs

The most frequent complications of the endovascular treatment of a cerebral AVM are hemorrhage and, less frequently, ischemia.

The main cause of hemorrhage seems to be an increase of the intra/perinidal vascular pressure after the embolization. Such increase of the vascular pressure responsible for bleeding may occur a) within the nidus (also in eventual arterial or venous pouch(es) including aneurysms and pseudo-aneurysms) and/or within the feeding pedicle(s) after inappropriate or involuntary occlusion of the AVM tributary venous drainage (without concomitant complete occlusion of the lesion itself which remains totally or partially permeable), b) within the adjacent normal arteries after the occlusion of the AVM allowing hyperemia of normal brain (so-called normal pressure breakthrough) and c) within a patent prenidial aneurysm after the occlusion of the nidus and/or of the AVM venous drainage. Hemorrhage may also be caused by progressive venous thrombosis due to stasis caused by significant AVM occlusion.

In the large series of Picard et al. [33], some pre-embolization AVM features, observed in lesions in which hemorrhage occurred after embolization, were the presence of steal phenomena (87%), multiple feeding arteries (100%), supratentorial location (100%), lobar topography (87%), a compact aspect of the nidus (93%), venous ectasia (53%) and venous stenosis (47%). Immediately after the endovascular treatment, angiographic findings which seemed to have a high amount of injected glue, significant venous embolization and persistent venous stagnation in and/or around the nidus. [33].

In the series of Deruty et al. [9] an occlusion of the main venous drainage could be demonstrated in 4 out of 5 cases of hemorrhage due to embolization. However, the hemodynamic mechanisms responsible for post-embolization hemorrhage remain not fully understood. Henkes et al. [16] measured the intra-arterial pressure in the AVM feeding pedicles using a microcatheter that had been placed for subsequent embolization treatment (201 measurements were performed in 95 patients before and after the embolization procedure). Their results showed a direct relationship between pressure changes and degree of embolization; however, the pressure changes after the embolization were relatively small. Then the Authors concluded that these pressure modifications are unlikely to be the direct cause of procedural bleedings. In addition to hemodynamic mechanisms, bleeding can be due to mechanical causes including vessel damage or perforation during the navigation (more often caused by the micro-guide) and rupture of the nidus or of a feeding pedicle at microcatheter removal after glue injection. However, in some cases, an AVM hemorrhage after endovascular treatment occurs without any identifiable causative factor [43]. As in our case, bleeding usually occurs in the early period after embolization. Picard et al. [33] reported 15 cases (3.05%) of hemorrhagic complications after AVM embolization, 3 patients bled during or immediately after embolization, 2 patients within 24 hours and 10 patients within 3 days after the procedure; in most cases (73%), bleeding was intraparenchymal. Deruty et al [9] reported 5 cases of hemorrhagic complication: 3 bleedings occurred during the endovascular procedure while the other 2 patients bled at day 2 and day 5 after embolization. In the series of Meisel et al. [30] and Steiger et al. [42], all embolization related hemorrhages occurred within 24 hours after embolization.

The ischemic complications in AVM endovascular treatment include embolic or thrombotic events due to catheterization and/or microcatheterization, anterograde glue injection into normal pedicles, retrograde glue injection into normal pedicles by reflux and retrograde thrombosis in the feeding arteries [9, 21]. After injection of the embolization material, the microcatheter can remain glued or trapped into the AVM [5]. In this case the microcatheter is left within the nidus and the feeding artery. As also demonstrated in experimental studies [22, 32], persistence of a microcatheter within a vessel is usually well tolerated especially if associate to pertinent pharmacological therapy; however, this event can be responsible for rarer ischemic complications.

In our case, we did not observed any glue migration into the AVM venous drainage and, despite a mild resistance during the microcatheter retraction, we had no elements in favor of the fact that it was glued within the AVM. However the microcatheter broke during retraction and we thought that the remained distal fragment at level of the MCA bifurcation could have caused an ischemic complication. In addition, we had no immediate angiographic signs evocative of a bleeding. The more likely hypothesis for the formation of the large hematoma was visualized on MR and CT scans is the possibility of vessel wall tearing of the AVM feeding pedicle associated to the rupture of the weakened microcatheter when the latter was being retrieved.

Hemorrhage appears the most frequent and serious complications in the endovascular treatment of
a cerebral AVM [20, 33, 35, 44]. Our complication had a terrible outcome and was responsible for the death of the patient.

In a paper published in 2001 [33], Picard et al. reported that, in their series, hemorrhage after embolization affected 15 (3.04%) patients out of 492 cases of totally or partially treated cerebral AVMs. Four patients were asymptomatic. Among the remaining 11 symptomatic patients, 6 were operated in order to evacuate an intracerebral hematoma; outcome was good in 7 patients, fair/poor in 2 (0.4% morbidity) and 2 patients died (0.4% mortality). Picard et al. [33] also reviewed the literature from 1972 to 1997 reporting hemorrhage after AVM embolization. Among 1206 patients with an AVM embo-

lized using different embolic agents, there were 58 (4.8%) cases of post-embolization bleeding. This review included the large series of Vinuela et al. [46] in which 11 patients out of 405 (2.7%) presented with hemorrhagic events after the AVM endovascular treatment. Frizzel et al. [11] reviewed the morbidity and mortality associated with the embolization of 1246 brain AVMs reported in the literature. Permanent morbidity was 9% before 1990 and 8% since 1990. Mortality associated with the AVM embolization was 2% before 1990 and 1% since 1990. Jafar et al. [20] published results in 20 patients who underwent AVM embolization using glue in order to facilitate surgical resection. Endovascular complications included immediate and delayed hemorrhage in 3 patients (15%) and transient ischemia in one patient (5%); there were no embolization-related deaths. In 1996, Deruty et al. [9] reported a high percentage of complications after AVM embolization: 10 out of 40 patients (25%). The mechanism of complication was hemorrhage in 5 cases and ischemia in the other 5. Complications were minor in 5 patients (12.5%), responsible for a deficit in 2 (5%) and death in 3 patients (7.5%). All deaths were attributable to hemorrhage due to the embolization procedure. In the same year, Wikholm et al. [48] reported results in 150 patients who underwent AVM embolization. The incidence of severe complications after embolization was 6.7% (10 patients) and the procedural mortality was 1.3% (2 patients). Consequently, the combined rate of death and complications affecting lifestyle was 8.0%, equal to approximately 3.2 years of natural history. Also in 1996, Gobin et al. [12] reported a series of 125 patients with AVM treated by embolization. Embolization resulted in a morbidity rate of 12.8% (16 patients) and a mortality rate of 1.6% (2 patients). The hemorrhage rate for partially embo-

lized AVMs was 3% per year. Consequently, the Authors concluded that the risk of hemorrhage from the residual nidus after partial embolization is comparable to the natural history of untreated AVMs. More recently, Kwon et al. [23] and Han et al. [13] reported that palliative endovascular treatment even increases the risk of future hemorrhage. On the contrary, Meisel et al. [29, 30] studied the yearly hemorrhage incidence rate after partial targeted glue embolization. In all untreated patients of their series this rate was 8.9%. In the subgroup of patients who underwent partial embolization, the observed annual rate was respectively 5.2% before and 3.6% two years after the start of treatment. The Authors concluded that
CONCLUSION

Evaluation of the natural history of brain AVMs shows that these lesions frequently have a poor or fatal outcome justifying a therapeutic approach. The risk of annual bleeding of a brain AVM is estimated from 2 to 4%. Morbidity from AVM rupture bleeding is estimated from 13% to 50% with a risk of mortality reported from 3 to 30% [2, 4, 14, 17, 31, 45].

Embolization is considered by most authors as the first step in the management of these lesions allowing, in some cases, complete AVM occlusion [6, 12, 20, 24, 30, 33, 43, 46]. There is also an important role for embolization before radiosurgery and surgical approach in order to make AVM exclusion possible and safer. Preoperative embolization, diminishing size and flow to an AVM, may also reduce intraoperative blood loss and operative time [20, 47]. However, despite improvement in techniques and understanding of these lesions, endovascular treatment of a cerebral AVM remains a challenging and delicate procedure. After AVM endovascular treatment, permanent morbidity risk is reported in 0.4% to 12.5% of patients and mortality in 0.4% to 7.5% [9, 11, 12, 20, 21, 30, 33, 42, 46]. More recently, AVM embolization–related permanent morbidity has been estimated to occur in 9% of patients and death in 2% [44]. However, we deal with statistical risks and the individual ratio risk/benefit is nowadays impossible to estimate. Hopefully in the future, experience and advances in knowledge and technical aspects could allow a more precise assessment of the therapeutic risk in each particular case. Hemorrhage appears to be the most frequent and serious complication in the endovascular treatment of a brain AVM. Severe hemorrhagic complications during or after embolization may also occur in AVMs without prior bleeding. In these cases, the treatment is performed in order to protect the patient of a potential risk, thus a severe or fatal complication is hardly accepted. The report of this case contributes to remind all of us involved in the management of brain AVMs that, even in centers with experience in the management of these lesions, AVM embolization can be followed by a poor and even fatal outcome. This risk must be always carefully considered and discussed between the medical team, the patient and its family for planning the AVM endovascular treatment.

REFERENCES


