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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 artérioveineuse 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 MAVs reste 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 sévère. 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 artérioveineuse 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

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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 Arterio-Venous 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 arteriolo-venular 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 positioned 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 mydriase 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-