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Case Report

Radionecrosis as a Late Complication of Gamma Knife Treatment of Brain Arteriovenous Malformation- A Case Report

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Abstract

We present a man with an arteriovenous malformation treated by medical therapy, embolisation, radiosurgery and neurosurgery and follow up the succes of the treatment through a fifteen year life period. It is also discussed about radionecrosis as a late gamma knife surgery complication.

Keywords: *Areteriovenous Malformation; Embolisation; Gamma Knife; Necrosis*

Introduction

Arteriovenous malformations (AVM) are abnormal connections of blood vessels bypassing the brain tissue and directly diverting blood from arteries to the veins [1]. It is estimated that the prevalence varies between 10 and 18 per 100.000, with an incidence between 1.1 to 1.3 per 100.000 person in a year [2]. It is still unknown why they occcur, but it is presumed that most of them are of congenital origin, either there are some reports of „de novo" brain arteriovenous malformations which occurs after tumor resection [3]. As they can occur anywhere within the brain like in frontal, parietal, temporal or occipital lobe, in the cerebellum, the brainstem, or in the ventricules, they can produce symptoms related to different brain functions depending on the location [4]. In more than 50% of patients with an AVM they have been recognized after an intracerebral haemorrhage [5]. In 20 - 25% of AVM they can present as focal or generalized seizures and in some cases they can bring to localized pain due to the increased blood flow around the AVM [6].

There are several potential treatment options for brain arteriovenous malformations. The main goal of the treatment is to prevent potential haemorrhage. The treatment possibilities include medical therapy to regulate the blood pressure, surgery, or interventional therapy consisting of embolisation-interventional neuroradiology/endovascular neurosurgery and gamma knife surgery/stereotactic radiosurgery, which lately in more and more cases replaces the traditional surgical resection [7].

By using gamma knife surgery there are several possible complications like local loss of hair in superficial lesions, local brain swelling on treated site, local necrosis, visual loss and deafness are [8]. In those cases a surgical removing of the necrotic brain lesion is needed to be done [9].

Case Report

We present a 60 years old patient with AVM located in the left frontal lobe. The first symptom was epilepsy which manifested as a grand mal attack in the year 2005., at his age of 46. He had no other neurological symptoms or deficits. The treatment started with antiepileptic drugs and the brain neuroimaging confirmed an AVM. Firstly an endovascular embolisation was performed in January 2006. with a partial result due to present anatomical difficulties which enabled the procedure to be totally succesful (Figure 1). The final part of internal carotid artery and the A1 part of the cerebral anterior artery were of specific anatomical configuration so it was not possible to bring the microcatheter to the right possition for embolisation. The medial part of AVM was vascularized from the right and the left anterior cerebral arteries, and the lateral part of AVM from the left medial cerebri artery. After completing the procedure the patient was taking only antiepileptics (carbamazepin) and antihypertensives.

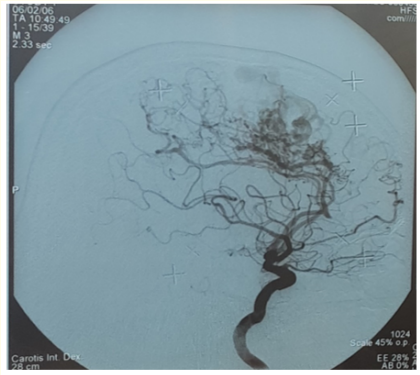

Figure 1: *DSA imaging after the first embolisation done in January 2006. with a partial result.*

One month after embolisation, gamma knife surgery by prescription dose of 14 Gy to prescription isodose 50 % was performed to reduce the mass effect of AVM. In the next few years the patient was regulary controlled clinically, and by brain magnetic resonance/ angiography and digital subtraction angiography (DSA) in 2006. and in 2010. year. Magnetic resonance (MR) imaging performed for several times showed volume regression of AVM (Figure 2). In 2010. year a retreatment with embolisation was done. In 2013. the MR angiography showed the obliteration of AVM. In November 2013. DSA confirmed the obliteration of the nidus and a fusiform expansion of the left medial cerebral artery was found.

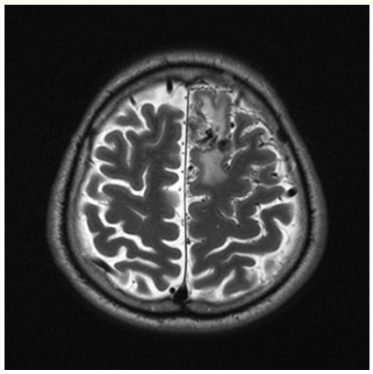

Figure 2: *MR brain imaging in 2010. showing the volume regression of AVM.*

On MR images in 2014. a development of brain oedema was evident. In 2015, ten years after the AVM was diagnosed and treated by embolisation (2006., 2010.) and by gamma knife therapy (2006), an intracerebral haemorrhage appeared in the left frontotemporal lobe as seen on figure 3. The digital subtraction angiography (DSA) showed incipient signs of brain vessels radionecrosis and an intensive antiedematous therapy was performed (Figure 4). In the next seven months the patient was treated conservative by taking corticosteroids, antihypertensive drugs and antiepileptics. In January 2016. it came to a rehemorrhagisation. CT scanning and brain MRI showed a haemorrhagia with great oedema and a contralaterall mass effect with a subfalx incipient herniation. The antioedematous therapy started immediatelly but gave a very poor result. A constant cooperation with a neurosurgeon brought us to conclusion that a neurosurgery has to be done. The AVM and the necrotic brain mass which appeared to be a late complication of brain radiation, was removed in February 2016. and the patient recovered well. The latest neuroimaging on figure 5 and 6 confirmed the good recovery.

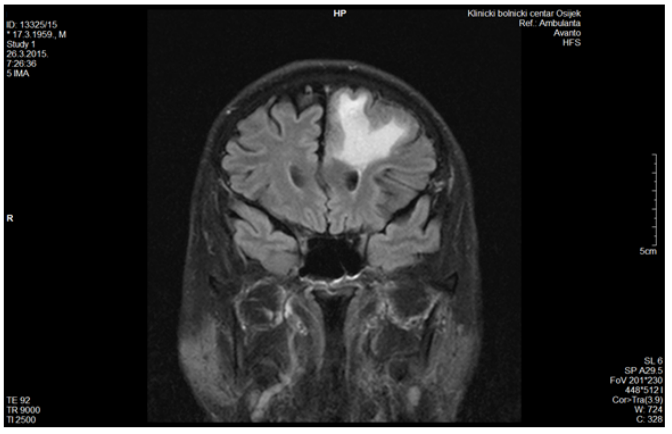

Figure 3: *Brain magnetic resonance in 2015. showing intracerebral haemathoma in the left frontotemporal lobe.*

Figure 4: *DSA in 2015. showing the initial necrosis of brain vessels after radiosurgery.*

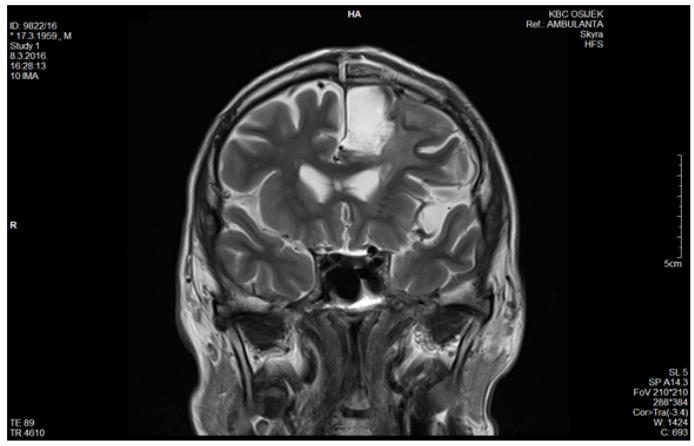

Figure 5: *Rezidual gliosis after neurosurgery treatment of left frontotemporal lobe with slight oedema.*

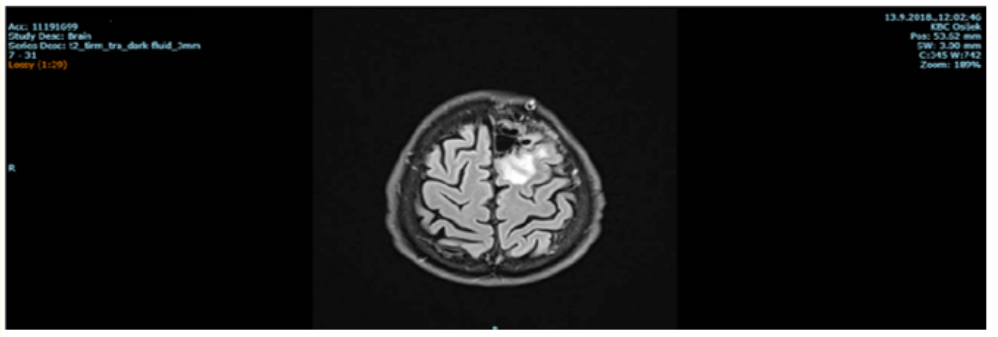

Figure 6: *Brain magnetic resonance imaging showing porencephalic cyst and gliosis of the surrounding tissue after neurosurgery.*

Discussion

Some authors suggest bevacizumab should be used in treating radionecrosis after radiosurgery of a solitar brain metastasis, but there is a lack of information whether it can be used in treating radionecrosis which is a consequence of a gamma knife surgery of brain AV malformation [10].

Hyperbaric oxygen therapy has been reported as a treatment option of radionecrosis and symptomatic brain oedema after stereotactic radiosurgery of AVM [11]. In most of cases when a symptomatic radionecrotic mass developes, an operative treatment is unavoidable [3].

In our patient the clinical corse suggested a medical therapy to avoid the epileptic seizures. An interventional embolisation was done in January 2006. and one month later a gamma knife therapy. A retreatment of embolisation was done in 2010. In the next five years the patient was stable untill 2015. when it came to a brain haemorrhage. The neuroimaging (DSA) confirmed incipient signs of vessel radionecrosis and after the second brain haemorrhage which was firstly treated conservative, an open surgical treatment had to be done. Due to extensive physical therapy it came to a good recovery of the left hemiparesis and the seizures were captured by using antiepileptic

drugs. The patient is now feeling good, has no headaches, but mild cognitive and speech impairment is present as well as a mild left side hemiparesis.

Some authors also presented a good recovery after a surgical removal of the necrotic brain mass which brought to immediate clinical improvement in cases with no corticosteroid response [12]. In our patient, the recovery was also good, and we concluded that the neurosurgical treatment after all previous procedures was the only treatment option [9].

AVM may present with various symptoms sometimes even uncommon as recurrent positional vertigo, which is described by some authors [13]. The symptoms depend on the brain region the AVM is located. In our patient the first manifestation was an epileptic seizure due to the cortical irritation. The possible treatments of AVM also vary from case to case but in rare cases embolisation brings to full recovery because it rarely occludes the nidus totally [14].

In treating complex brain AVM with embolisation the outcome is often only partial so GKS is commonly used afterwards [15]. Some authors questioned the embolization prior to stereotactic radiosurgery in comparisson to RS alone and they concluded that the risk of late haemorrhagisation is not correlated to prior embolisation before RS [16]. Comparing the projected risk of surgery with no intervention Morgan., *et al.* concluded that it is better to do surgery in expert centers than to remain untreated [17].

Our patient experienced almost all possible methods for treating AV malformations. Firstly he was treated medicamentously and afterwards by embolisation performed as an endovascular procedure by a neurosurgeon. After that a gamma knife surgery was performed, but due to the appearance of radionecrosis as a late radiosurgery complication, an open neurosurgical treatment was performed to remove the necrotic brain mass. It is known that radionecrosis is a late complication of radiotherapy with limited therapeutic options, but latelly bevacizumab is desribed as a possible treatment option [18].

Sharon., *et al*. showed two cases of delayed radionecrosis after stereotactic radiosurgery suggesting that residual iron deposits after prior haemorrhagisation could be a radiation sensitizer contributing to the adverse event years after the procedure [19]. In our case radionecrosis appeared prior of haemorrhagisation and is not correlated to possible iron deposits.

A medical algorythm for treating AV malformations does not exist and the treatment depends on a variety of different factors. The treatment with bevacizumab and hyperbaric oxygen therapy is still not used routinely in this specific and quite rare disorder.

Conclusion

Ten years after the AVM was diagnosed and treated by endovascular surgery, an intracerebral haemorrhage appeared. The digital substraction angiography (DSA) showed a radionecrosis of brain vessels so an intensive antiedematous therapy was performed. The antioedematous therapy was not succsesfull, so a surgery was done to remove the AVM and the necrotic brain mass which appeared as a late complication of brain radiation. Today no algorithm exists for treating AVM, because each AVM is somewhat specific and each patient requires individualized approach. Future studies are necessary in order to achieve longer alleviation of neurological symptoms, especially after brain radiation.

We conclude that one treatment is not superior above other, but they are most succesful when used in combination.

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