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It's Time for the Next Paradigm Shift in Pulmonary Arterial Hypertension Management?

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Introduction

In ARUBA, the annual hemorrhage rate of unruptured AVMs was found to be 2.2% [1], while the re hemorrhage rate according to other studies is 6%–15% for the first year [2-6]. It is well known that the presence of intracranial arterial aneurysms (AA) in patients with AVMs, that reach nearly 3-58% according to different data [7- 15], is associated with an increased risk of hemorrhage [2, 6, 7, 12, 14, 16]. Study of unruptured AVMs between 1974 and 1985 at the Mayo Clinic revealed the risk of hemorrhage among patients with a coexisting of unruptured AVM and AA was 7% at 1 year compared with 3% among those with an AVM alone. This higher risk of hemorrhage persisted at 5 years in patients with AVMs and AAs (7%/year) compared with patients with AVMs alone (1.7%/year) [1].

Treatment of patients with AVMs and AA is challenging and controversial. But targeting of both lesions at the same sitting may be preferable [9]. Improving of endovascular treatment modalities including guide-wire-directed microcatheter delivering liquid adhesive embolic agents and detached coils have considerably improved the efficacy of AVM and aneurysms occlusion. We have analyzed 421 cases of AVM from our institute over the a 12-year-period and we suggest that endovascular technique can be an effective treatment option to minimize the complications in patients with AVM associated with AAs.

Clinical Material and Methods

Patient population

Four hundred twenty one patients with angiographically verified cerebral AVMs that were treated in SI «Scientific-Practical Center of Endovascular Neuroradiology NAMS of Ukraine» between 2004 and 2016 were evaluated. Among these patients, 91 (21.6%) patients were diagnosed with AVMs and accompanying intracranial AAs. All this patients were included in this analysis. There were 47 (51,6 %) women and 44 (48,4 %) men, ranging from 10 to 62 years with a mean age of 42 years.

Classification of the associated arterial aneurysms

Cerebral AAs associated with AVMs were divided according to Perata et al [17]. Based on hemodynamic dependence of AA and AVM and their anatomical correlation into four groups: intranidal AAs, flow-related (distal) AAs of vessels supplying the AVMs, flow-related (proximal) AAs of the circle of Willis origin of an artery supplying to the AVM, AAs of remote artery that was not involved in the AVM supply. Saccular AAs arising along the course of arteries that eventually supplied the AVM were classified as flow related. Flow-related AAs were subclassified as proximal if they were located on the supraclinoid Internal Carotid Artery (ICA), the circle of Willis, the Middle Cerebral Artery (MCA) up to and including the primary bifurcation, the Anterior Cerebral Artery (ACA) up to and including the ACoA, or the Vertebrobasilar Trunk (VT). All other flow-related AAs were subclassified as distal [18]. Simple arterial ectasias, infundibulae, venous pouches, and variceal dilations were excluded. In 91 (21.6%) of these 421 patients, we observed 67 (59,8 %) intranidalAAs, 25 (22,3 %) flow-related AAs of vessels supplying the AVMs, 17 (15,2 %) AAs of the circle of Willis origin of an artery supplying to the AVM and 3 (2,7 %) AAs of remote artery that was not involved in the AVM supply.

Clinical courses

According to clinical and imaging findings, we determined the courses of AAs. Hemorrhage was defined as a symptomatic event, when we found blood on imaging or in the cerebrospinal fluid after LP. To define the origin of the hemorrhage, we used multiplanar reconstructions and angiography techniques in CT, DSA or MRI. We also consider anatomic relationship of the AVM and the AA

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and intraoperative findings.

Treatment

In ruptured AA, we usually focused on the AA, aiming for it occlusion within 24 hours. In unruptured AA, we developed an individualized treatment strategy, bearing in mind that there are reports of AA shrinkage after successful AVM treatment. Endovascular treatment was performed under general anesthesia after administration of 10,000 IU of heparin and included AA coiling or embolization with liquid embolization agents. Superselective microcatheter was cannulated of arteries feeding the AVM and coils, Embolin, NBCA or Onyx18 were used to occlude the AAs. In selected cases, the balloon remodeling technique or endovascular stent placement was necessary.

Embolization of the nidus was usually performed using Embolin, NBCA and Onyx under general anesthesia. Neurological outcomes were based on the Glasgow Outcome Scale (GOS) at discharge.

Statistical analysis

All statistical analyses were performed using commercially available software (SPSS 22.0. Trial version, IBM, 2014). P-values < 0.05 were considered significant.

Results

112 AA were detected in 91 patients with AVM. Multiple AA were present in 13 (14,2 %) patients (Figure 1). Eight (8,7%) AVMs were located infratentorial and 83 (91, 3%) were supratentorial. Hemorrhage was the most frequent presenting symptom in 63 patients (69, 2 %). We found that 43 (68, 2 %) hemorrhagic cases were caused by AVM rupture with intranidal AAs, 12 (19,1 %) cases by rupture of AAs of vessels supplying the AVMs, 7 (11, 2 %) - AAs of the circle of Willis origin of an artery supplying to the AVM, 1 (1,5 %) - remote AAs that was not involved in the AVM supply. The mean diameter of all AAs was 4.2±3.1 mm (1–23 mm, median 3.8 mm). A hemorrhagic presentation was more frequent in the subgroup with intranidal AAs 43/67 (64, 1%) than the subgroup with flow-related AAs 17/42 (40,4%) (chi-square test, $p < 0.02$).

Twenty eight patients had no history of hemorrhage, and 24 (26, 3 %) of these suffered seizure and the other 4(4, 5 %) complained headache, dizziness, vertigo (where routine CT, MRI reveled AVMs, and DSA - coexisting AA).

All patients were treated by endovascular approach for a AVM-related AAs by using coils, NBCA and Onyx.21 AAs (18,8 %) were coiled (Figure 1-3) and other 82 (73, 2 %) flow-related and intranidal AAs were embolized with Embolin, NBCA and Onyx18 (Figure 4). There were eight patients (8 %) with AVM, with nine associated AAs who did not receive any treatment for their AAs, two of them with 3AAs because of terminal state after hemorrhage. 3 patients had fusiform flow-related arterial AAs, that were left for observation, 1 patient had proximal flow-related arterial ACoA microaneurysm with a complicated afferent vessels anatomy that was hard to occlude but it regress after subtotal AVM treatment (Figure 5), and 2 patients reject any surgery. Two patients (2,1%) died after AAs rupture. None patient died after procedure. Excellent or good outcomes (Glasgow Outcome Scale ≥4) were observed in 81 (89 %) patients at discharge. Unfavorable outcomes (Glasgow Outcome Scale 1–3) had 10 (11 %) patients at discharge.

Follow-Up

All 91 patients had clinical follow-up from 1 month to 12 years starting at the date of the first documentation of the AVM (with CT, MRI, DSA). Eighty (87,9 %) had angiographic follow-up after embolization, starting at the time of the first embolization session, ranging from 1 to 19 months (mean, 6.2 months). None of the coiled aneurysms reanalyzed. One (0, 89 %) hemodynamically related AAs regressed during follow-up, and none of the residual AAs rupture during the follow-up period. Five patients showed de-novo AA formation after subtotal AVM obliteration, all of them in cases of flow-related AAs.

Discussion

There is no constant consensus on any question about AAs associated with AVMs, except it worsen the clinical course. First of all, pathogenesis is still not well known, but during the last decade development of IAs are frequently associated with hemodynamic factors dictated by the presence of shunting in the AVM nidus. This theory is supported by the observation that most AAs are located on proximal arteries hemodynamically connected to the nidus [13]. Proximal flow-related AAs frequently occur at bifurcations, distal flow-related AAs frequently occur along the course of the feeding artery pedicle, not related to bifurcations, and often have irregular shapes and a wide neck [14].

The incidence of AAs associated with AVMs varies widely among different studies (2.7%–58%) [4]. In a latest meta-analysis the incidence of AAs was reported to be 18% [6]. Endovascular surgery, microsurgery and radiosurgery are available treatment options. However, all have pros and cons. Our strategy, in which endovascular treatment is the primary option, is based on our possibilities and experience. Even if the AVM can't be completely cured with the endovascular approach, we believe that embolization is indicated in all patients with a high risk of rupture or re-rupture. In patients who have a hemorrhage at presentation, the first issue is to determine the site. If the AA is suspected, it is treated first with either glue or coils; the choice depends on characteristics such as location and size. If the nidus of the AVM is responsible for the bleeding, the treatment is aimed primarily at the AVM.

If the source of the bleeding cannot be established radiologically, we focus our treatment on the AA. In patients who have not had a hemorrhage, our treatment strategy depends upon many factors, including aneurysmal location, size of the nidus, and topography.

For accurate evaluation of AAs associated with AVMs it is important to understand the nature of arterial pseudoaneurysm, that was first described by Garcia-Monaco et al in 1993 [5]. Arterial pseudoaneurysms commonly originate from small perforating arteries or choroidal branches and indicates the point of rupture. They often show progressive enlargement on repeat angiography. If the source of the hemorrhage is a pseudoaneurysm, then either direct treatment or close imaging follow-up should be utilized. If the pseudoaneurysm is treated, the AVM can then be treated electively later because the source of hemorrhage has been secured. Endovascular techniques offer a particular advantage when intravascular access can be safely achieved in the vicinity of the pseudoaneurysm, given that most pseudoaneurysms are located on perforating arteries, which can be difficult to reach with an operation [13]. In our series, spontaneous aneurysmal regression was observed in 1 case and remained unchanged on follow-up angiograms. We also found 5 (5, 4 %) cases of de-novo aneurysm formation, but in contrast to Thompson et al. [15], they were silent [15].

According to our experience, treatment options for IAs associated with AVMs should be weighed according to risk-to-benefit ratio on the expertise of the institution and physicians. All treatment modalities have their own complication rates i.e. surgery (29%; range, 1.5%–54%), endovascular treatment (25%; range, 7.6%–55%), and radiosurgery (13%; range, 0%–63%) [17]. And according to ARUBA study of previously unruptured AVMs has 30.7% complication rate for all treatment modalities) [10].

Limitations of all current study including our own are retrospective character, patients selection problems and referral bias. But it seems that the main question about IAs associated with AVMs is long term follow-up that will compare different kinds of treatment and of course natural history of this association.

Conclusions

The type of endovascular treatment depend on the site of the AA and its relationship to the nidus of the AVM. The main policy is to treat the symptomatic lesion first. Discovery of AA during AVM evaluation should turn it in the therapeutic focus. The method of choice is the simultaneous AA and AVM occlusion and elimination of associated aneurysms is principal.

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