CASE REPORT

Delayed rupture of a basilar artery aneurysm treated with coils: Case report and review of the literature

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KEYWORDS
Aneurysm; Rupture; Complications; Embolization; Intracranial

Summary Delayed rupture of a previously unruptured cerebral aneurysm after uneventful saccular coil packing is rare, particularly when the quality of aneurysm occlusion is appropriate (neck remnant or total occlusion). The present report describes the case of a 70-year-old woman with an incidentally detected, asymptomatic, small basilar tip non-thrombosed aneurysm who experienced rupture of the aneurysm 2 years after coiling. Cerebral angiography taken on the day of rupture revealed only small recanalization of the aneurysm neck with no dome-filling. This is the first report of delayed rupture due to minor recurrence of a previously unruptured small asymptomatic cerebral aneurysm after saccular coil packing. A literature review of 26 reports of late bleeding after coil embolization of previously unruptured cerebral aneurysms showed that dome-filling after coil embolization, symptomatic aneurysms and large/giant aneurysms all increase the risk of delayed rupture in previously unruptured aneurysms after saccular coil packing.

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Introduction

The efficacy of endovascular coil embolization in the management of unruptured cerebral aneurysms has been characterized in several large studies [1,2], and delayed rupture of a previously unruptured cerebral aneurysm after uneventful saccular coil packing is a rare event [3,4].

Case report

A 70-year-old woman was incidentally diagnosed with an asymptomatic basilar tip aneurysm on magnetic resonance imaging (Fig. 1) and magnetic resonance angiography (MRA). Her history was notable for hypertension, hyperlipidemia and coronary artery disease (treated with aspirin 100 mg daily), but she had no history of subarachnoid hemorrhage.
Transarterial coil embolization of this unruptured basilar tip aneurysm (diameter: 7.4 mm; Fig. 2 A–D) was performed, and 95% neck-remnant occlusion of the aneurysm was achieved using a balloon-neck remodeling technique (Fig. 2E). Computed tomography performed the next day showed no subarachnoid hemorrhage (Fig. 3).

MRA at 6 months after the first intervention revealed complete obliteration of the aneurysm (Fig. 4). However, after 18 months, MRA showed small recanalization at the neck of the aneurysm (Fig. 5).

Then, at 22 months after the initial treatment, the patient presented with subarachnoid hemorrhage (Hunt and Hess grade 3, Fisher group 3; Fig. 6A). Cerebral angiography demonstrated a small recurrence at the level of the neck of the basilar tip aneurysm with no other intracranial aneurysm (Fig. 6B–D). On the day of onset, transarterial coil embolization was performed, using a balloon-neck remodeling technique. During the chronic phase of subarachnoid hemorrhage, endovascular treatment was performed with a stent-assisted coil embolization technique, deploying two enterprise stents (Cordis Neurovascular, Miami Lakes, FL, USA) in a Y configuration and inserting coils through the stent mesh (Fig. 6E). Triple antiplatelet therapy, comprising aspirin 100 mg daily, cilostazol 200 mg daily and ozagrel 160 mg daily, was maintained during the perioperative period of stent-assisted coiling. The patient experienced neurological recovery up to a modified Rankin Scale score of 1.

Discussion

Delayed rupture after saccular coiling

In the published literature, there are 26 reports of late bleeding after coil embolization of previously unruptured aneurysms (Table 1) [5–21]. Ten were symptomatic and only one was asymptomatic at the time of initial presentation. In the other 15 cases, the presence or absence of symptoms was not reported. However, two of these 15 cases

Figure 1   Brain MRI (T1- and T2-weighted images) taken prior to initial embolization. The basilar tip aneurysm is seen with no thrombus or perianeurysmal edema.
Table 1  Cases reported in the published literature of delayed rupture of previously unruptured cerebral aneurysms after coil embolization.

<table>
<thead>
<tr>
<th>Authors, year</th>
<th>Aneurysm location</th>
<th>Size</th>
<th>Results of embolization&lt;sup&gt;a&lt;/sup&gt;</th>
<th>Occlusion rate at time of rupture</th>
<th>Embolization to rupture time lag</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Malisch et al., 1997 [7]</td>
<td>—</td>
<td>Giant</td>
<td>Residual aneurysm</td>
<td>—</td>
<td>12–20 months&lt;sup&gt;b&lt;/sup&gt;</td>
<td>Partially thrombosed aneurysm</td>
</tr>
<tr>
<td>Hodgson et al., 1998 [8]</td>
<td>Middle cerebral artery</td>
<td>Small</td>
<td>Residual aneurysm</td>
<td>Residual aneurysm</td>
<td>18 months</td>
<td>Partially thrombosed aneurysm</td>
</tr>
<tr>
<td>Eskridge et al., 1998 [9]</td>
<td>—</td>
<td>Giant</td>
<td>—</td>
<td>—</td>
<td>31–90 days</td>
<td>Partially thrombosed aneurysm</td>
</tr>
<tr>
<td>Murayama et al., 1999 [10]</td>
<td>Posterior communicating artery</td>
<td>Large</td>
<td>Residual aneurysm</td>
<td>Residual aneurysm</td>
<td>91–365 days</td>
<td>Partially thrombosed aneurysm</td>
</tr>
<tr>
<td>Birchall et al., 2001 [13]</td>
<td>Basilar tip</td>
<td>Giant</td>
<td>Residual aneurysm</td>
<td>—</td>
<td>8 months</td>
<td></td>
</tr>
<tr>
<td>Ng et al., 2002 [14]</td>
<td>—</td>
<td>—</td>
<td>Residual aneurysm</td>
<td>—</td>
<td>7 months</td>
<td></td>
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<tr>
<td>Brilstra et al., 2004 [16]</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>2 months</td>
<td></td>
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<tr>
<td>Gonzalez et al., 2004 [17]</td>
<td>—</td>
<td>Giant</td>
<td>Residual neck</td>
<td>—</td>
<td>8 months</td>
<td></td>
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<tr>
<td>—</td>
<td>Large</td>
<td>Complete occlusion</td>
<td>Residual aneurysm</td>
<td>13 months</td>
<td></td>
<td></td>
</tr>
<tr>
<td>van Rooij et Sluzewski, 2006 [18]</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td></td>
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<tr>
<td>Standhardt et al., 2008 [19]</td>
<td>Vertebrobasilar junction</td>
<td>Giant</td>
<td>Residual neck</td>
<td>Residual aneurysm</td>
<td>4 years</td>
<td>Partially thrombosed aneurysm</td>
</tr>
<tr>
<td>Piotin et al., 2010 [20]</td>
<td>Basilar tip</td>
<td>Giant</td>
<td>Residual aneurysm</td>
<td>Residual aneurysm</td>
<td>21 months</td>
<td>Partially thrombosed aneurysm</td>
</tr>
<tr>
<td>Pyysalo et al., 2010 [21]</td>
<td>—</td>
<td>Large</td>
<td>Residual aneurysm</td>
<td>—</td>
<td>7 years</td>
<td></td>
</tr>
<tr>
<td>Present case</td>
<td>Basilar tip</td>
<td>Small</td>
<td>Residual neck</td>
<td>Residual neck</td>
<td>22 months</td>
<td></td>
</tr>
</tbody>
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<sup>a</sup> Angiographic results at initial embolization and time of rupture classified according to the Montreal Scale.

<sup>b</sup> Used because no precise description given in report.

—: Data not described in report.
developed new compressive symptoms in the postoperative follow-up period prior to rupture of the aneurysm. Symptomatic aneurysms, which suggest recent growth and wall instability [22], could be associated with an increased risk of delayed rupture.

In addition, 16 delayed ruptures occurred in large and giant aneurysms, while only two were associated with small aneurysms. A high aspect ratio (dome-to-neck ratio) correlates with aneurysm rupture [23]. Large and giant aneurysms tend to have a high aspect ratio and should therefore receive particular attention even after coiling.

All reported cases of delayed rupture of previously unruptured aneurysms after saccular coil packing were seen in aneurysms with dome filling (residual aneurysm) [24,25], and 17 of the 26 aneurysms were incompletely occluded (residual aneurysm) at the time of initial embolization. Three aneurysms were subtotally occluded (residual neck) at the time of initial embolization; however, coil compaction and dome filling of the aneurysm were revealed on follow-up cerebral angiography. No descriptions regarding the degree of occlusion were given in the other six cases, and no cases (except for the present case) ruptured without dome filling. Rupture occurs preferentially at the site of the aneurysm dome [23]; thus, a completely occluded or neck-remnant-occluded aneurysm may be considered relatively safe for the prevention of aneurysm rupture.

Figure 2  Preoperative three-dimensional digital subtraction angiography, anteroposterior view (A) and rostrocaudal view (B). Right vertebral angiography, anteroposterior view (C) and lateral view (D), shows a basilar tip aneurysm 7.4 mm in diameter. Right vertebral angiography just after the initial saccular coiling shows 95% neck-remnant occlusion of the aneurysm (E).
Figure 3 Computed tomography performed on the day after initial embolization shows no intracranial hemorrhage.

**Figure 4** Follow-up magnetic resonance angiography done 6 months after initial embolization shows complete occlusion of the aneurysm.

**Figure 5** Follow-up magnetic resonance angiography performed 18 months after initial embolization shows small recanalization at the neck of the aneurysm.

Delayed rupture after flow-diversion treatment

Flow diverters are increasingly being used for the treatment of wide-necked cerebral aneurysms, and delayed rupture of the treated unruptured aneurysm after flow-diverter implantation has been reported in some studies [26]. The mechanisms of delayed rupture after such treatment are probably different from those following saccular coiling. In our review of the literature, the time delay between saccular coiling and rupture ranged from 4 hours to 7 years. Most ruptures (18 out of 20) happened more than 3 months after embolization (Table 1). In contrast, Kulcsar et al. [22] reported that delayed rupture after flow-diverter treatment occurred more frequently within 3 months of treatment: there were ten early (<3 months) ruptures and three late (3–5 months) ruptures among 13 delayed-rupture cases reported in patients with previously unruptured cerebral aneurysms treated with a flow diverter.

Implantation of a flow diverter led to significant flow changes around the aneurysm; however, inflow jets were still present immediately after treatment in most patients with delayed rupture [22]. Computational fluid dynamics analysis of cerebral aneurysms that ruptured after the deployment of flow diverters revealed that placement of
the device could increase intra-aneurysm pressure, thereby potentially causing rupture of the aneurysm [27].

Another potential mechanism of delayed rupture involves post-flow-diversion thrombosis of the aneurysm sac. Thrombosis of the sac can activate an inflammatory reaction at the aneurysm wall, which can result in weakening of the cerebral aneurysm [28] even though thrombus formation is the first step towards permanent aneurysm repair through cicatrisation and reverse remodeling of the vessel wall. Dual antiplatelet therapy after flow-diversion treatment could also play a role in delayed rupture. Most patients treated with flow diverters were still taking both aspirin and clopidogrel at the time of rupture [22].

**Treatment of recanalized aneurysms**

Our present case is the first report of delayed rupture due to the minor recurrence of a previously unruptured small asymptomatic cerebral aneurysm after saccular coil packing. It indicates that while the risk of bleeding from such a minor recurrence in a previously unruptured cerebral aneurysm is far less than that for a previously ruptured cerebral aneurysm [12], bleeding can still occur. Therefore, a repeat operation should be considered when recurrence of the aneurysm is detected.

Several treatment strategies can be used for reoperation of aneurysm recurrence, including balloon-neck remodeling, stent-assisted coiling and flow diverters; however, treatment may be difficult in cases with anatomically complex aneurysms, including a small recurrence and/or a wide neck. The balloon-neck remodeling technique has equivalent safety and achieves better occlusion compared with the standard coiling technique [29]. This means that the wider use of the remodeling technique can be proposed not only for recanalized unruptured aneurysms, but for ruptured/re-ruptured aneurysms as well. Stent-assisted coil embolization is another helpful technique for re-embolization of a minor recurrence in a wide-necked aneurysm [30], and was used to achieve total occlusion in the present case. However, aneurysm stent placement is
associated with a higher mortality compared with coiling with or without balloon-neck remodeling [20]. Accordingly, the wider use of stents is probably not to be recommended [29]. A flow diverter is yet another option for the initial treatment and retreatment of anatomically complex aneurysms; however, treatment using flow diverters can be difficult in the context of dual antiplatelet therapy, particularly for ruptured/re-ruptured aneurysms.

Conclusion

The risk of bleeding of a minor recurrence of a previously unruptured cerebral aneurysm after saccular coil packing is extremely low. The presence of dome-filling after coil embolization, symptomatic aneurysms and large/giant aneurysms all increase the risk of delayed rupture in such coil-packed, previously unruptured aneurysms at usually more than 3 months after coil embolization.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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