Multiple Dural Arteriovenous Fistulas

Satoshi USHIKOSHI, Yoichi KIKUCHI*, Kiyohiro HOUKIN**, Hisatoshi SAITO, and Hiroshi ABE**

Sapporo Azabu Neurosurgical Hospital, Sapporo; Departments of *Radiology and **Neurosurgery, University of Hokkaido School of Medicine, Sapporo

Abstract

Four patients with multiple intracranial dural arteriovenous fistulas (DAVFs) at separate sites were treated by endovascular techniques (transarterial and/or transvenous embolization), surgery (excision or isolation), radiotherapy, or combinations, according to the pathophysiological condition. All lesions in two patients were obliterated completely without neurological deficit. There were residual fistulas after the treatment in two patients, but these were low-grade lesions without retrograde cortical venous drainage, and marked clinical improvement was obtained. Planning of treatment strategies for multiple DAVFs requires careful analysis of the venous drainage from the affected sinuses and cerebral hemodynamics.

Key words: dural arteriovenous fistula, multiple lesions, endovascular therapy

Introduction

Intracranial dural arteriovenous fistulas (DAVFs) are currently considered to be acquired abnormal arteriovenous connections located within the wall of a dural sinus.2,6,8,17 Widely spreading contiguous fistulas, such as those involving the transverse-sigmoid sinuses, are not infrequent, but multiple DAVFs at separate sites are relatively rare.1,5,6,10,12,13,15,16,20 The incidence of multiple lesions was 6.7% in a large number of patients with DAVF.11 We treated four patients with multiple DAVFs over the last 12 years, and describe the pathogenesis and treatment strategies for multiple DAVFs.

Materials and Methods

The clinical presentation, radiological findings, treatment, and outcomes of the four patients with multiple DAVFs treated at our hospital were reviewed. Each lesion was classified according to the revised Djindjian classification4 based on the type of venous drainage (Table 1). Abnormalities of dural sinuses were also examined. Treatment strategies, including endovascular techniques, surgery, and radiotherapy, were determined individually based on the pathophysiology. Transarterial embolization was performed using polyvinyl alcohol (PVA) particles or n-butyl 2-cyanoacrylate (NBCA). Transvenous embolization was performed from the femoral vein, using platinum coils as the embolic materials.

Table 1 Classification of dural arteriovenous fistulas according to venous drainage

<table>
<thead>
<tr>
<th>Type</th>
<th>Venous drainage</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Into a sinus, with normal anterograde flow</td>
</tr>
<tr>
<td>II</td>
<td>Into a sinus with insufficient anterograde venous drainage and reflux</td>
</tr>
<tr>
<td></td>
<td>a: into sinus(es) only</td>
</tr>
<tr>
<td></td>
<td>b: into cortical vein(s) only</td>
</tr>
<tr>
<td></td>
<td>a + b: into sinus(es) and cortical vein(s)</td>
</tr>
<tr>
<td>III</td>
<td>Directly into a cortical vein, without venous ectasia</td>
</tr>
<tr>
<td>IV</td>
<td>Into a cortical vein, with venous ectasia</td>
</tr>
<tr>
<td>V</td>
<td>Into spinal perimedullary veins</td>
</tr>
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According to the revised Djindjian classification.4

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Author's present address: S. Ushikoshi, M.D., Department of Radiology, University of Hokkaido School of Medicine, Sapporo, Japan.
the dural wall of the sinus or stimulates the development of such shunts through the recanalization process. Recently, the importance of both sinus thrombosis and venous hypertension was demonstrated in the development of DAVF in animal models. Moreover, an animal experimental study showed that only venous hypertension, without thrombosis, can cause the development of arteriovenous shunts. Based on these experimental results and some histological evidence, the elevated venous pressure in a dural sinus or a vein is probably transmitted directly to the vasa vasorum-like capillaries and arterioles in the sinus or venous wall via a retrograde route, resulting in the formation of arteriovenous shunts. Sinus thrombosis may be involved by causing venous hypertension or activating angiogenetic factors during the recanalization process.

The pathogenesis of multiple DAVFs is difficult to determine, as these lesions may develop at different sites simultaneously by the same process, or the first DAVF may induce development of the others. Case 1 had a second DAVF in the previously occluded sinus, and located downstream to the first DAVF. Case 2 developed a DAVF following thrombosis of the superior sagittal sinus, located upstream to the
other two DAVFs. There are several explanations for the development of multiple DAVFs. The venous drainage due to an established DAVF may cause turbulent flow or stagnation in the distant venous sinus, resulting in thrombosis of the sinus and development of other DAVFs. Thrombosis of a dural sinus at several sites caused by, for example, hypercoagulative state may also lead to the development of multiple DAVFs. However, sinus thrombosis could not be demonstrated in Cases 3 and 4, although severe stenosis of both jugular veins was recognized in Case 4. Two interesting cases of development of new DAVF during the follow-up period for the initial DAVF have been reported.\textsuperscript{5} As only venous hypertension unaccompanied by sinus thrombosis can cause the development of DAVF, elevated sinus pressure caused by initial DAVF or some other pathogenesis can result in the formation of multiple new DAVFs at other sites.

Currently, endovascular techniques are the first
choice for most DAVFs.\textsuperscript{5,7,11,14,18} Transarterial embolization is very useful for reducing the shunt flow, but complete cure is usually difficult to attain due to the recruitment of other feeding vessels and recanalization of the embolized vessels when particulate embolic materials are used. Recently, transvenous embolization was proposed as an effective and curative treatment.\textsuperscript{7,14} However, venous infarction may occur after occlusion of dural sinuses or cortical veins which contribute to the normal venous drainage or are not exposed to the high venous pressure. The decision to perform transvenous embolization should be based on the patient’s clinical history and the risks of the treatment. From this point of view, grading systems for classifying DAVF have been proposed based on the venous drainage pattern.\textsuperscript{2,5,14} The most appropriate candidates for transvenous embolization are patients with cortical venous drainage and normal veins of brain tissue not draining into the affected sinus.\textsuperscript{2,5,7,11,14,18}

Patients with multiple DAVFs present with an even more complex state. Venous hypertension caused by one DAVF might be enhanced by the presence of the other DAVF. The treatment of multiple lesions must consider whether multiple sinus occlusion can be tolerated, especially when both the sagittal and the transverse-sigmoid sinuses, or the bilateral transverse-sigmoid sinuses are affected, as in our Case 4. Previously, three cases of combined sagittal and lateral sinus DAVF were treated with excellent clinical outcomes, and the effectiveness of staged deliberate occlusion of the affected sinuses was emphasized.\textsuperscript{13} Treatment of DAVF should attempt to decrease venous hypertension and improve the cerebral hemodynamics, not to obtain complete obliteration of the fistulas.\textsuperscript{1,13,15} Examination of the venous drainage from the affected sinuses and brain tissue is most important, and the appropriate strategy should be devised accordingly. Our and previous experience suggests that hazardous multiple DAVFs and clinical symptoms can be controlled successfully by staged obliteration of the fistulas. Changes in cerebral hemodynamics and clinical presentations should be investigated carefully at each step for determination of the next step of treatment.\textsuperscript{13} Persistent low-risk lesions without retrograde cortical venous drainage do not always require treatment, although the natural history of such lesions is not yet well known.

References

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Address reprint requests to: S. Ushikoshi, M.D., Department of Radiology, University of Hokkaido School of Medicine, North-15, West-7, Kita-ku, Sapporo 060-8638, Japan.