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## Treatment of Dural Arteriovenous Fistulae (dAVF's) at the Superior Sagittal Sinus (SSS) Using Embolisation Combined with Micro- or Radiosurgery

### Abstract

dAVF's at the SSS are extremely rare and usually present with intracranial haemorrhage (ICH) or a progressive neurological deficit. Due to their midline location and multiple arterial supply, endovascular treatment alone often fails in eliminating the fistula. Therefore, endovascular, combined with neurosurgical and/or radiosurgical treatment is often needed to cure the patient. We summarized our experience with three male patients over a ten-year period who suffered from dAVF's involving the middle and posterior third of the SSS. Two of them presented with an ICH during the clinical course. Despite multiple transarterial embolisations, complete fistula occlusion could not be achieved in any of them. Nevertheless, neurological symptoms improved in all cases. One patient refused further treatment and died six years later due to an ICH. Another patient was finally cured by microsurgical coagulation of the fistula. In the remaining patient stereotactic radiosurgery (SRS) was performed after nearly complete endovascular occlusion. We strongly recommend microsurgery and/or radiosurgery as adjunctive measures, if embolisation alone fails to eliminate these dangerous fistulae.

### Key words

Dural Arteriovenous Fistula · Superior Sagittal Sinus · Embolisation · Microsurgery · Stereotactic Radiosurgery · Gamma Knife

### Introduction

Intracranial dAVF's are rare and account for 10%–15% of all intracranial arteriovenous malformations [1–4]. Most frequently, they involve the transverse or cavernous sinus. Those located at

the SSS are extremely rare [1,3,4,5–7]. Their frequency is reported to be between 8%–13% of all dAVF's [1,4,6,8].

The etiology of these "spontaneous" fistulas is still controversial. Predisposing factors like head trauma, cerebral or peripheral infections, intracranial surgery and even hormonal disturbances are reported to play a role in the development of dAVF's [1,6,7,9,10]. Pathological studies revealed large thin-walled venous pouches in close proximity to small dural arteries and arterioles inside the normal dura mater. These venous pouches may rupture due to an arteriovenous pressure gradient and create an arteriovenous shunt that may grow because of the permanent pressure gradient [1,6,7,11].

Depending on venous drainage, dAVF's may present with benign symptoms like bruit or headaches, but also can cause fatal subdural, subarachnoid or intraparenchymatous haemorrhage [1,5,7,10,12,13].

Different forms of treatment include manual percutaneous compression, transvascular embolisation, radiosurgery and open surgery [3,5–7,10,13–16].

Because of their midline location and the multiplicity of arterial feeders associated with a critical venous drainage, SSS-fistulae have a reported rate of bleeding up to 40%. This percentage is higher than in dAVF's at other locations and therefore often unusual and more aggressive forms of treatment are required [6,7,10,15,17–19].

During a ten-year period we encountered three such rare lesions that could not be occluded by one form of treatment alone. In two of them endovascular treatment combined with micro- or

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### Bibliography

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radiosurgery was performed. The remaining patient died 6 years after partial endovascular occlusion of his fistula as he refused complementary surgical treatment.

## Case Reports

### Case 1

A 51-year-old male patient was admitted to another neurosurgical department because of sudden severe headache, diplopia and a pulsatile bruit. Computerized tomography (CT) revealed distended vascular channels adjacent to the SSS and no ICH or subarachnoid hemorrhage (SAH) could be seen. Angiography disclosed a dAVF involving the posterior part of the SSS and the confluens sinuum. In the medical history a viral infection with meningeal affection was found six months earlier.

After transferral to our institution, superselective angiography showed the dAVF to be supplied by both external occipital arteries (EOA), the left posterior auricular artery (PAA), both superficial temporal arteries (STA), both middle meningeal arteries (MMA) and tentorial branches of both internal carotid arteries (ICA). The fistula drained into the SSS, which appeared to be partly occluded by an intraluminal thrombus in its posterior part. The fistula was classified as a type IIa fistula, according to Cognard et al. [1]. After superselective catheterisation of the main feeders as close to the fistula as possible, multiple dural nidi were embolised, but the fistula could not be completely occluded in one session. Postoperatively, a discrete left-sided facial palsy occurred, whereas all other symptoms improved.

Three weeks after dismissal from our institution symptoms reoccurred. Angiography revealed that a new arterial supply from right-sided ECA branches had developed. Reembolisation was performed, but again total occlusion was not possible due to small remaining shunts from tentorial ICA branches.

Again symptoms improved, but reoccurred three weeks thereafter with the new symptom of a right-sided trigeminal hypoesthesia and a facial palsy. Embolisation was performed, but AV shunts coming from both ascendent pharyngeal arteries and the tentorial ICA branches remained. In the course of three embolisations, microcoils (Target Therapeutics, Fremont, CA) and particles (IVALON<sup>®</sup>, Ingenor, Paris, France) were used. Microsurgical disconnection of the SSS from its arterial supply was proposed, but the patient refused further therapy. A control angiography eight weeks later demonstrated an unchanged condition. Within the following six years neurological symptoms markedly improved. Hypoesthesia, diplopia and the bruit completely disappeared. The facial palsy improved with mild headaches remaining.

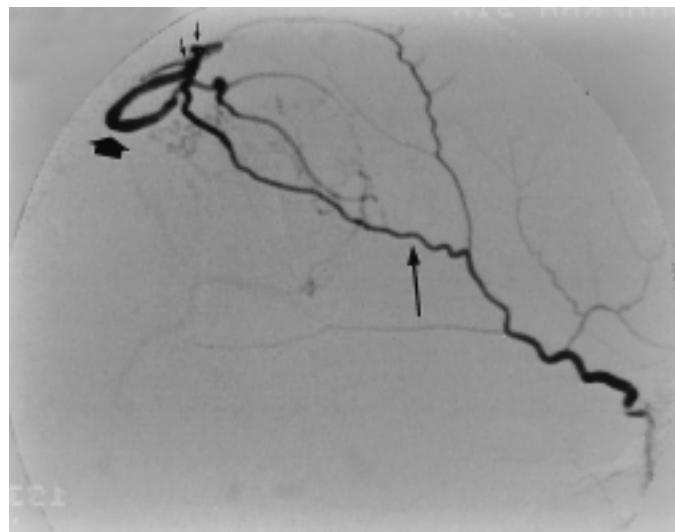
However, six years later the patient suffered from a large left-sided ICH that had to be evacuated in emergency at another neurosurgical department. On preoperative angiography a dAVF could not be detected. Two weeks later the patient rebled and finally died.

### Case 2

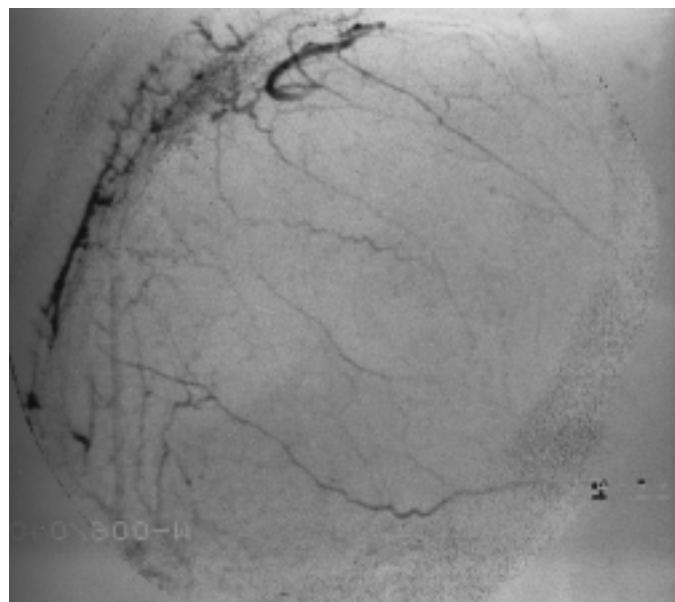
A 57-year-old male patient was admitted to another neurosurgical department because of sudden headaches, a hemihypoesthesia and severe weakness of the left arm. CT revealed an ICH in the

right motor strip. Magnetic resonance imaging (MRI) showed a cluster of distended vessels adjacent to the haemorrhage near the central sulcus. Angiographic studies including both ECAs indicated a dAVF located at the middle part of a completely patent unobstructed SSS. In the medical history head trauma, infection or surgery was absent.

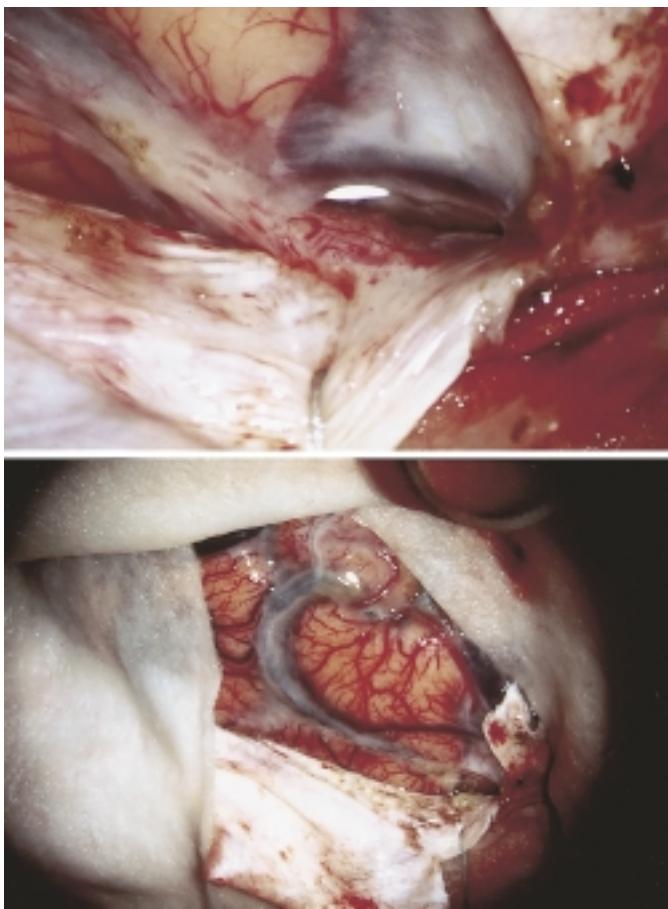
Superselective angiography at our institution showed a dAVF type III, that was fed by the both MMAs and the right STA filling up a pial, cortical, comma-shaped vein adjacent to the SSS, that retrogradely filled cortical veins including the anastomotic vein of Trolard (Fig. 1a). The fistula was embolised twice using the glue (N-butyl-2-cyanoacrylate, HISTOACRYL BLAU<sup>®</sup>, Braun Melsungen, Germany) and particles via both MMAs and the right STA. Postinterventional angiography revealed a persistent opacification of the comma-shaped pial vein via cutaneous transosseous EOA branches (Fig. 1b).



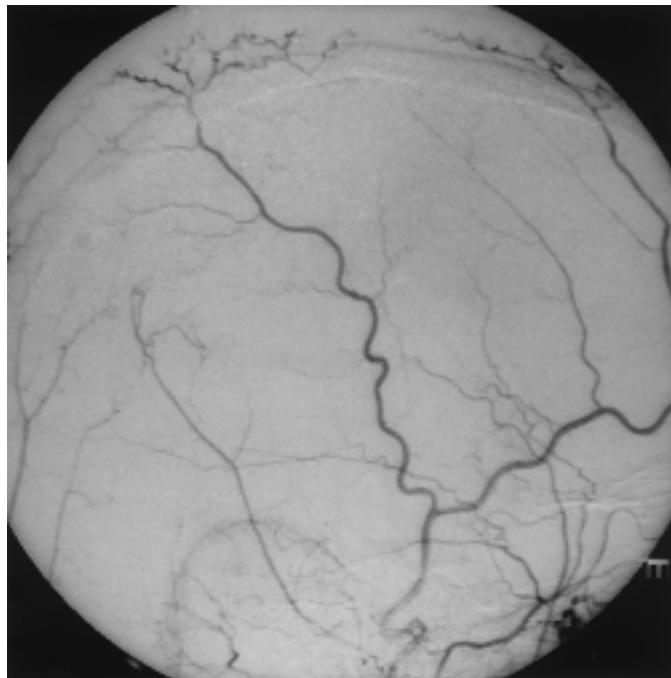
**Fig. 1a** Superselective angiogram of the right middle meningeal artery (MMA) (→) filling up a comma-shaped pial fistulous vein (→), lateral view.



**Fig. 1b** After embolisation, the vein is still fed by cutaneous, transosseous external occipital artery (EOA) branches, lateral view.



**Fig. 1c** Intraoperative view showing the pial, comma shaped draining vein with shunting arterioles on its wall at the entry point to the SSS before (above) and after coagulation (below).



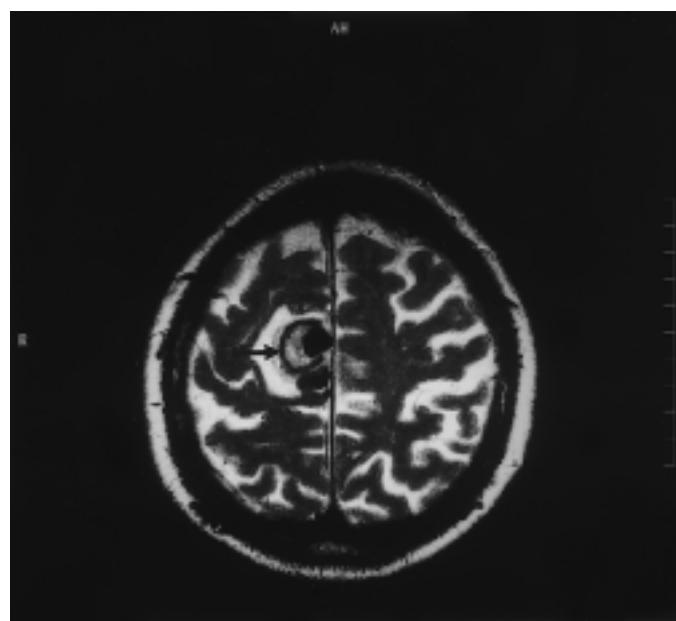
**Fig. 1d** Superselective angiography of the right external carotid artery, one year after operation shows complete elimination of the fistula, lateral view.

One week later, the fistula was explored via a small parasagittal craniotomy and occluded microsurgically. Intraoperatively, we found a partially thrombosed pial draining vein which was still red in its distal part due to shunting arterioles running in its thickened wall. The vein was coagulated and transected at its entrance point to the SSS, the adjacent dura was coagulated too. Excision of the adjacent dura was not performed (Fig. 1c). Within the following months the patient made a complete recovery. Follow-up angiography two years later showed no residual fistula and a patent SSS (Fig. 1d).

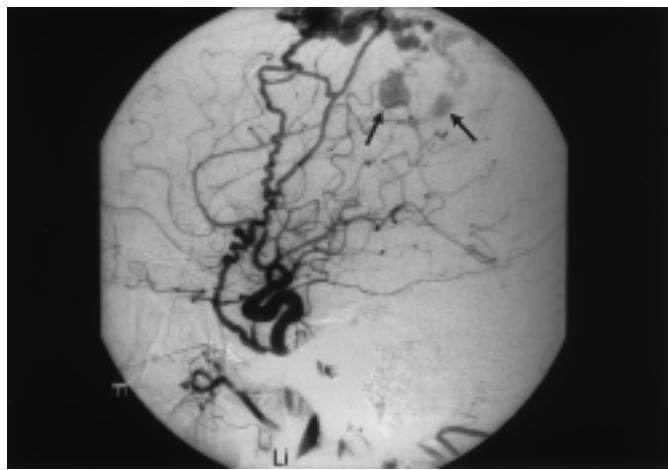
### Case 3

A 71-year-old male patient suffered from a sudden monoplegia of the left leg followed by a general seizure. At another institution MRI showed an ICH located at the right motor strip and venous varices (Fig. 2a). At our institution angiography revealed a type IV dAVF, with feeders coming from the anterior falx branch of the right ophthalmic artery, both STAs and both EOAs. The venous drainage occurred through cortical veins to the middle part of a patent SSS posterior to the fistula site with two aneurysmal dilatations (Fig. 2b, c). The anterior and middle part of the SSS appeared occluded by a thrombus.

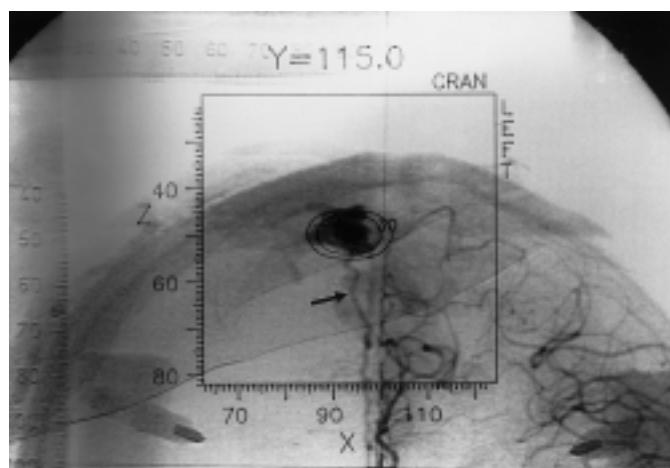
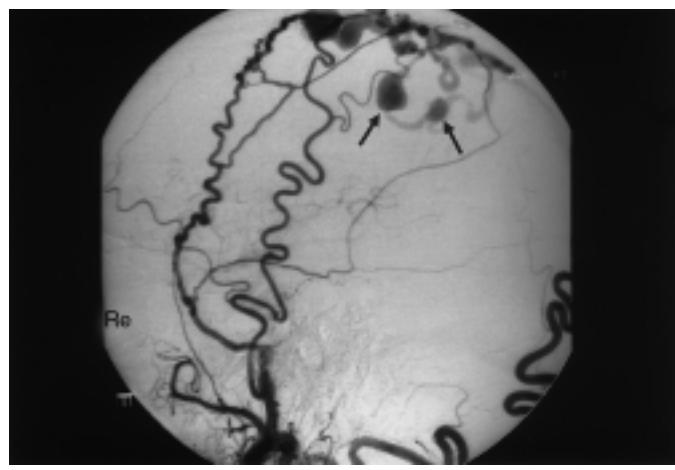
Medical history demonstrated no predisposing factors, but a recent myocardial infarction. Within two weeks we performed two transfemoral embolisations via both MMA's using glue. During the third intervention we performed surgical exposure of both STA's and both EOA's near the SSS for direct embolisation (Fig. 2d). A nearly total occlusion of the fistula was achieved, with only the anterior falx branch remaining, which fed the anterior part of the fistula (Fig. 2e, f). Due to his poor medical condition we decided to treat him one week later by stereotactic radiosurgery with the gamma-knife unit. The fistula site was covered with the 50% isodose and treated with 22 Gy using three 8 mm



**Fig. 2a** T<sub>2</sub>-weighted MRI, demonstrates an intracerebral clot adjacent to two venous varices (→), axial view.



Figs. 2b,c Angiography of the left common carotid artery (CCA) (b) and the right external carotid artery (ECA), (c), shows the fistula and the two venous aneurysms (→).



Figs. 2e,f Preradiosurgical angiogram reveals the residual fistula fed by the anterior falx artery (→), a/p-view (e) and lateral view (f).

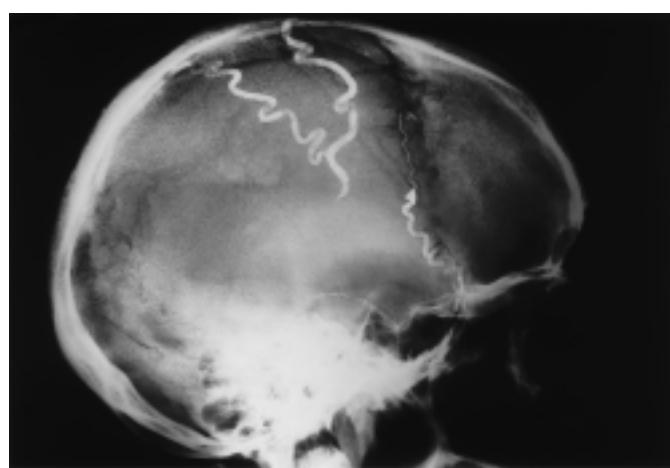
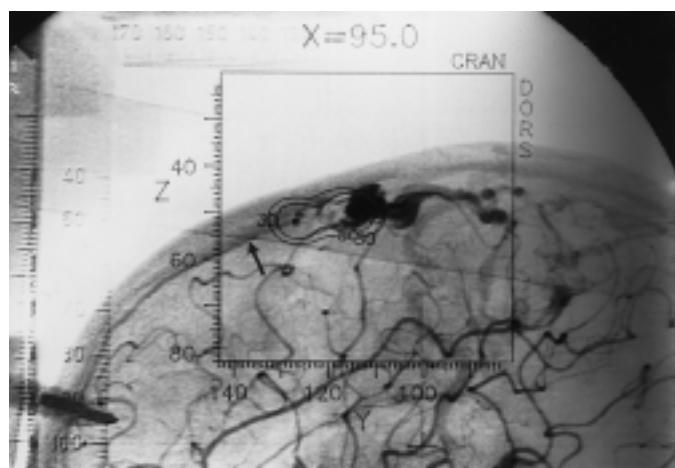


Fig. 2d Conventional X-ray of the skull after surgical exposure and embolisation via the right superficial temporal artery (STA) shows the cast in the artery and near the fistula site.

collimator helmets (Fig. 2e,f). During the following weeks the patient recovered completely.

## Discussion

Dural AVF's involving the SSS account for 8%–13% of all intracranial dAVF's [1–4,10]. Usually they occur with a female predominance at an age of 40 years and above [1,6,9]. Predisposing factors like head trauma, infection or intracranial surgery may lead to thrombosis of a cranial sinus, which is regarded to be the inciting factor in the development of a dAVF [6,7,9]. Due to progressive organisation of the thrombus inside the sinus by ingrowing arterioles and capillaries an arteriovenous communication develops in the sinus wall itself. Therefore, dAVF's are often associated with occlusion or stenosis of the fistula carrying sinus. Our current study involves three male patients, with an age ranging typically from 59 to 71 years. In the medical history, only one patient had already had a viral meningitis half a year earlier. Angiographically an SSS occlusion was seen in case 3 and an SSS stenosis in case 2. In case 1 a patent, unobstructed sinus was demonstrated on control angiograms two years after treatment.

The clinical behaviour of a dAVF seems to be at least partly explained by its location [5]. For example, dAVF's located at the tentorial incisura have a higher incidence of presenting with an aggressive clinical course than dAVF's located at the cavernous sinus or at the transverse/sigmoid sinus, although no location is immune to ICH [5]. In dAVF's involving the SSS the incidence of bleeding is reported to be up to 40% [6,7,10,12,18]. All of our three patients presented with an aggressive neurological course, two of them with the initial symptoms of an ICH. The remaining patient experienced two ICH's, that finally led to a fatal outcome.

Another point to be stressed is the pattern of venous drainage, that highly correlates with the clinical course [1]. Cognard et al introduced a reviewed classification for dAVF's based on the fistulous venous drainage [1]. According to them, type I dAVF's drain into a sinus with antegrade venous outflow. A type IIa has a reflux into the sinus, a type IIb presents with reflux into cortical veins, and a type IIa+b with both, reflux into the sinus and into cortical veins. Type III is characterised by direct cortical venous drainage without draining into the sinus. A type IV has additional venous ectasias and type V drains exclusively via spinal veins. The incidence of an intracerebral haemorrhage increases with the type of dAVF. In our patients we encountered one type IIa, a type III and a type IV fistula. The type III and the type IV dAVF initially presented with an ICH. We assume that the IIa fistula type developed into a higher grade lesion during the clinical course, which eventually led to two ICH's. In Cognard's series the bleeding rate for type IIa was 0%, for type III 40% and for type IV-fistula 66% [1].

Different single or combined treatment modalities have been suggested for dAVF's, depending on location, neurological course and venous drainage pattern of the fistula. Forms of treatment include percutaneous compression of feeding arteries, transarterial or transvenous embolisation, microsurgery or stereotactic radiosurgery (SRS). In the literature, transvascular embolisation followed by transcranial microsurgery or radiosurgery is favoured, if total occlusion is not possible [3,6,7,13,15,20,21,22]. Only few reports exist concerning the treatment of dAVF's involving the SSS. At that site most authors recommend transvascular embolisation using particles or glue, either alone or in combination with open microsurgery, or SRS [6,7,10,17,19]. Van Halbach et al. reported on two series consisting of seven and ten dAVF's involving the SSS [6,10]. A complete radioanatomical cure by arterial embolisation alone was achieved in 50%. 30% of the SSS fistulae were treated by intraoperative exposure of the feeding arteries for direct embolisation. The remaining 20% of the patients needed additional open surgery to eliminate the fistula. In two cases resection of the involved sinus was necessary. Pierot et al. described a dAVF at the SSS accompanied by an occluded sinus [19]. After the failure of transarterial embolisation, transvenous embolisation and surgical sinus skeletonisation, the fistula was finally occluded by surgical exposure of the SSS, direct puncture and embolisation of the entire SSS with free spirals. Cloft et al. described a successful transvenous coil embolisation of an SSS fistula [17]. Picard et al. regard transarterial embolisation as the method of choice, sometimes with the necessity of percutaneous puncture of the feeding arteries. If embolisation fails they recommend surgery [7]. In our patients we used various methods including the combination of transarterial embolisation, embolisa-

tion after surgical exposure of the feeding arteries, microsurgical resection and stereotactic radiosurgery.

Even though total occlusion of dAVF's can be achieved by the different means listed above, it is not expected to happen after application of one treatment method alone. Especially dAVF's at the SSS are difficult to cure completely compared to dAVF's at other sites, due to their midline location, their bilateral, multiple, and often cutaneous arterial supply as well as the involvement of a mostly critical venous cortical drainage. For this reason, transarterial embolisation alone is a sufficient method to cure the fistula in only 50% of SSS dAVF's when compared to dAVF's located at other sites [6,10,23]. The same is true for open surgery alone with its inherent danger of the occurrence of disastrous intraoperative arterial bleeding or of severe neurological postoperative deficit due to the compromise of the venous drainage of both hemispheres when the SSS has to be resected [10,24]. Combined treatment modalities are therefore indicated [1,5,6,7,10,13].

In our first patient multiple transarterial embolisations with coils and particles reduced the size and flow of the dAVF. Even though occlusion of many arteriovenous shunts involving a long distance of the SSS occurred, a total fistula occlusion was not achieved. Unfortunately, further surgical treatment was refused. In the following years the fistula did not occlude spontaneously, despite excellent clinical recovery of the patient, who was nearly without symptoms for the following six years. However, he finally died shortly after having experienced two ICH's.

In cases 2 and 3, who both presented with ICH's, the treatment consisted of multiple arterial embolisations followed by microsurgery in one case and by stereotactic radiosurgery in the other. Despite moderately sized ICH's having occurred, both patients recovered completely and finally had an excellent neurological outcome. Follow-up angiography showed complete eradication of the fistula in the patient who was operated on. The patient who underwent radiosurgery has a limited angiographic follow-up study. To demonstrate complete occlusion this fistula is expected to disappear over the period of one year, as SRS already proved an excellent tool in the treatment of dAVF's with a minimal risk of recurrent ICH in the waiting period for total occlusion, provided the flow through the fistula has been markedly reduced by previous embolisations [13,15,20,25].

As our experience with three SSS fistulae has shown total angiographic occlusion should be the aim of therapy in each case. There is no proof that partial fistula occlusion has any positive influence on the grave natural history of these aggressive dAVF's. Because of the extraordinary arterial supply and involvement of the SSS over a long distance, one cannot assume spontaneous fistula occlusion after partial embolisation. In addition, partial arterial occlusion can lead to recanalisation of embolised vessels or recruitment of other feeding arteries less accessible to embolisation. Therefore, additional forms of treatment are needed. At first we recommend transarterial embolisation with glue or particles until the size of the dAVF is reduced. For the remaining feeders, surgical exposure for direct puncture near the fistula might be necessary. By that means the flow through the dAVF is subsequently decreased and the venous drainage of the brain is improved. Furthermore, the number of blood units is reduced in

case open surgery becomes necessary. If the patient is in a poor medical condition, which precludes open surgery, transarterial embolisation followed by radiosurgery may be an effective alternative tool to finally obliterate the fistula.

## References

- <sup>1</sup> Cognard C, Gobin YP, Pierot LP, Bailly AL, Houdard E, Casasco A, Chiras J, Merland JJ. Cerebral dural arteriovenous fistulas: Clinical and angiographic correlation with a revised classification of venous drainage. *Radiology* 1995; 194: 671–680
- <sup>2</sup> Newton T, Cronqvist S. Involvement of the dural arteries in intra-cranial arteriovenous malformations. *Radiology* 1969; 90: 27–35
- <sup>3</sup> Bavinzski G, Richling B, Killer M, Gruber A, Levy D. Evolution of different therapeutic strategies in the treatment of cranial dural arteriovenous fistulas – Report of 30 cases. *Acta Neurochir (Wien)* 1996; 138: 132–138
- <sup>4</sup> Brown Jr RD, Wiebers DO, Nichols DA. Intracranial dural arteriovenous fistulae: angiographic predictors of intracranial hemorrhage and clinical outcome in nonsurgical patients. *J Neurosurg* 1994; 81: 531–538
- <sup>5</sup> Awad IA, Little JR, Akrawi WP, Ahl J. Intracranial dural arteriovenous malformations: factors predisposing to an aggressive neurological course. *J Neurosurg* 1990; 72: 839–850
- <sup>6</sup> Halbach VV, Higashida RT, Hieshima GB, David FD. Endovascular therapy of dural fistulas. In: Vinuela F (ed). *Interventional neuroradiology: Endovascular therapy of the central nervous system*. New York: Raven, 1992: 29–50
- <sup>7</sup> Picard L, Bracard S, Moret J, Pez A, Giacotte HL, Roland J. Spontaneous dural arteriovenous fistulas. *Seminars in interventional radiology* 1987; 4: 219–241
- <sup>8</sup> Gaston A, Chiras J, Bourbotte G, Leger JM, Guibert-Tranier F, Merland JJ. Meningeal arteriovenous fistulae draining into cortical veins. *J Neuroradiology* 1984; 11: 161–177
- <sup>9</sup> Chaudahary MY, Sachdev VP, Cho SH, Weitzner Jr I, Puljic S, Huang YP. Dural arteriovenous malformation of the major venous sinuses: An acquired lesion. *AJNR* 1982; 3: 13–19
- <sup>10</sup> Halbach VV, Higashida RT, Hieshima GB, Rosenblum M, Cahan L. Treatment of dural arteriovenous malformations involving the superior sagittal sinus. *AJNR* 1988; 9: 337–343
- <sup>11</sup> Roland J, Bernard C, Bracard S. Microvascularisation of the intracranial dura mater. *Surg Radiol Anat* 1987; 9: 43–49
- <sup>12</sup> Lasjaunias P, Chiu M, Brugge KT, Tolia A, Hurth M, Bernstein M. Neurological manifestation of intracranial dural arteriovenous malformations. *J Neurosurg* 1986; 64: 724–730
- <sup>13</sup> Lewis Al, Tomsick TA, Tew Jr JM. Management of tentorial dural arteriovenous malformations: transarterial embolisation combined with stereotactic radiation or surgery. *J Neurosurg* 1994; 81: 851–859
- <sup>14</sup> Barnwell SL, Halbach VV, Higashida GB, Wilson GB. Complex dural arteriovenous fistulas: Result of combined endovascular and neurosurgical treatment in 16 patients. *J Neurosurg* 1989; 71: 352–358
- <sup>15</sup> Link MJ, Coffey RJ, Nichols DA, Gorman DA. The role of radiosurgery and particulate embolisation in the treatment of dural arteriovenous fistulas. *J Neurosurg* 1996; 84: 804–809
- <sup>16</sup> Sundt Jr TM, Piepgras DG. The surgical approach to arteriovenous malformations of the lateral and sigmoid dural sinuses. *J Neurosurg* 1983; 59: 32–39
- <sup>17</sup> Cloft HJ, Kallmes DF, Jensen ME, Dion JE. Percutaneous transvenous coil embolisation of a type 4 sagittal sinus dural arteriovenous fistula: Case report. *Neurosurgery* 1997; 41: 1191–1193
- <sup>18</sup> Kurl S, Saari T, Vanninen R, Hernesniemi I. Dural arteriovenous fistulas of the superior sagittal sinus: case report and review of the literature. *Surg Neurol* 1996; 45: 250–255
- <sup>19</sup> Pierot L, Visot A, Boulin A, Dupuy M. Combined neurosurgical and neuroradiological treatment of a complex superior sagittal sinus dural fistula: Technical note. *Neurosurgery* 1998; 42: 194–197
- <sup>20</sup> Guo WY, Pan DH, Wu HM, Chung WY, Shiao CY, Wang LW, Chiou HJ, Yen MY, Teng MM. Radiosurgery as a treatment alternative for dural arteriovenous fistulas of the cavernous sinus. *AJNR* 1998; 19: 1081–1087
- <sup>21</sup> Martin NA, King WA, Wilson CB, Nutik S, Carter LP, Spetzler RF. Management of dural arteriovenous malformations of the anterior cranial fossa. *J Neurosurg* 1990; 72: 692–697
- <sup>22</sup> Mayfrank L, Reul J, Hoffmann B, Bertalanffy H, Spetzger U, Gilsbach JM. Microsurgical interhemispheric approach to dural arteriovenous fistulas of the floor of the anterior cranial fossa. *Minim Invas Neurosurg* 1996; 39: 74–77
- <sup>23</sup> Halbach VV, Higashida RT, Larsen DW, Dowd CF, McDougall CG, Hieshima GB, Wilson CB. Treatment of dural arteriovenous fistulas. In: Maciunas RJ (ed). *Endovascular neurological intervention*. Park Ridge: AANS, 1995: 217–246
- <sup>24</sup> Jaeger R. Observations and resections of the superior longitudinal sinus at and posterior to the rolandic inflow. *J Neurosurg* 1951; 8: 103–109
- <sup>25</sup> Chandler Jr HC, Friedman WA. Successful radiosurgical treatment of a dural arteriovenous malformation: case report. *Neurosurgery* 1993; 33: 139–142