

# Gamma Knife Surgery for the Treatment of Spontaneous Dural Carotid-Cavernous Fistulas

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## Abstract

Endovascular treatment for a spontaneous dural carotid-cavernous fistula (CCF) is an established treatment, but stereotactic radiosurgery might provide a less-invasive alternative in selected cases. Four women aged 67 to 79 years (mean 72.0 years) with spontaneous dural CCFs presented with chemosis or bruit. Angiography revealed arteriovenous fistulas in the cavernous portion. Three cases were Barrow type D and one was type B converted from type D. Stereotactic gamma knife surgery was performed with a marginal dose of 13–15 Gy and a maximum dose of 26–30 Gy with a volume from 824 to 1755 mm<sup>3</sup>. The target point of radiosurgery for type D CCFs was the compartment of the cavernous sinus supplied by multiple feeders from the external carotid artery. All patients responded favorably to the treatment, with improvement of symptoms beginning after 1 to 3 months. Angiography confirmed the complete disappearance of the CCFs in all patients. There were no recurrences, and the follow-up period was 14 to 32 months (mean 24 months). No significant side effects were observed. Stereotactic radiosurgery is a useful method to treat CCFs and is indicated for elderly patients, low-flow CCFs, and cases in which endovascular treatment has failed.

Key words: spontaneous carotid-cavernous fistula, stereotactic radiation therapy, elderly patients

## Introduction

Transvenous or transarterial embolization for dural carotid-cavernous fistula (CCF) is an established method, but such endovascular treatment carries the risk of permanent complications in difficult cases.<sup>6,7)</sup> Radiosurgery for CCFs is controversial, but provides a less-invasive treatment for patients who meet the required indications. We treated four elderly patients with low-flow dural CCFs by stereotactic gamma knife surgery as the first treatment in three cases.

## Materials and Methods

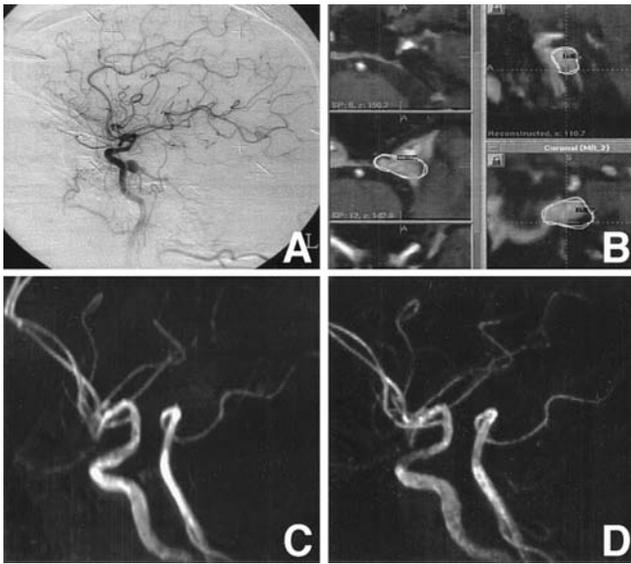
This study included four women aged 67–79 years (mean 72 years) with CCFs discovered after findings

of chemosis or bruit. Angiography revealed arteriovenous fistulas (AVFs) in the cavernous portion, and the fistulas were classified according to Barrow's classification.<sup>2)</sup> Carotid compression was performed in all patients but failed to provide the desired results.<sup>11)</sup> Digital subtraction angiography (DSA) superimposed on magnetic resonance (MR) imaging was used to identify the shunt point.

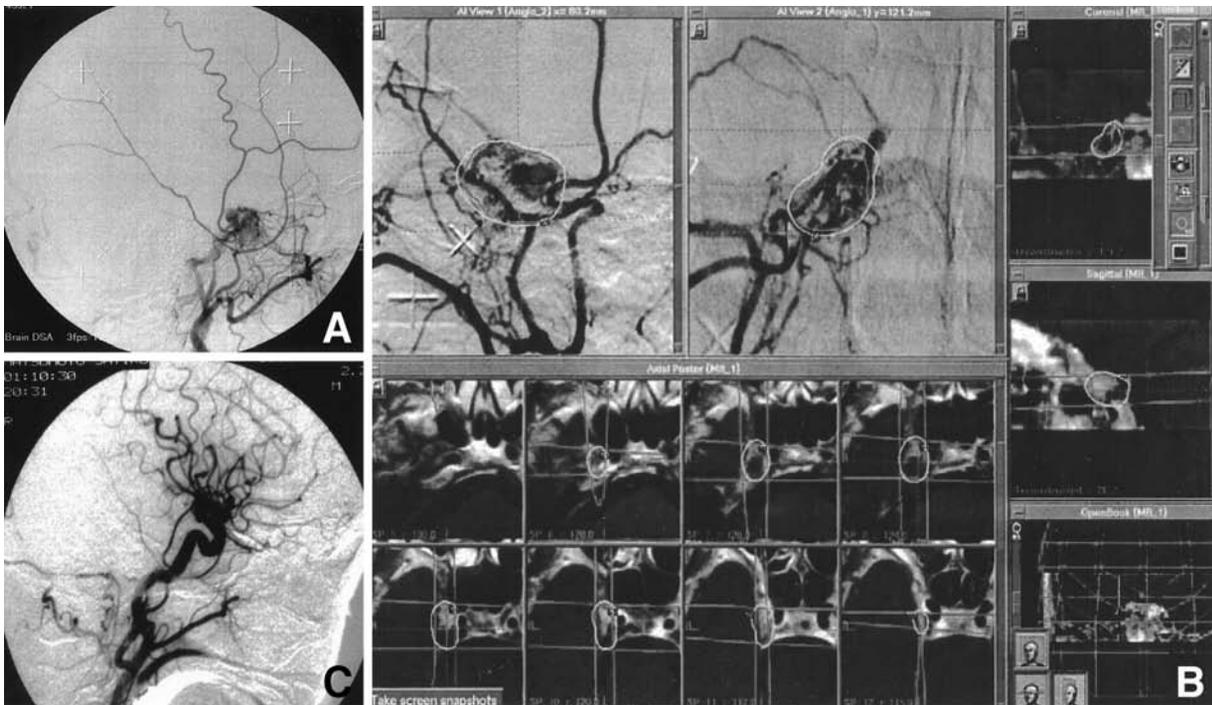
Gamma knife radiosurgery was performed at 2–9 months (mean 4 months) after the onset of the symptoms. All fistulas located along the wall were included in the radiation target. In particular, the target point of radiosurgery for type D CCFs included the compartment of the lateral wall and inferior wall of the cavernous sinus supplied by multiple feeders from the external carotid artery (ECA). Feeding arteries and drainage veins distal to the sinus

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**Fig. 1** Case 1. A: Left carotid angiogram showing a dural arteriovenous fistula of the cavernous sinus converted from type D to type B after embolization of the external carotid artery branches. B: Digital subtraction angiograms superimposed on magnetic resonance (MR) images showing the dose planning for radiosurgery. C, D: Follow-up MR angiograms showing persistence of the fistula after 6 months (C), but disappearance after 2 years (D).

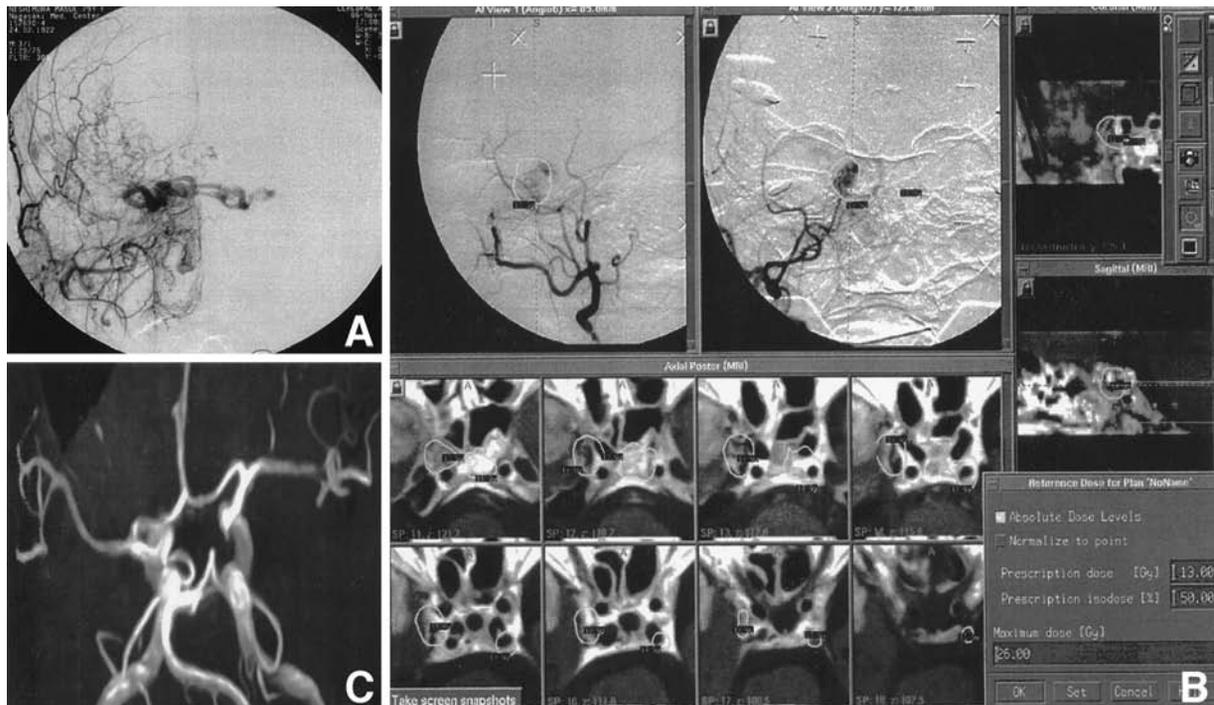


**Fig. 2** Case 2. A: Right carotid angiogram showing a cavernous dural arteriovenous fistula classified as type D. B: Digital subtraction angiograms superimposed on magnetic resonance images showing the dose planning for radiosurgery. C: Right carotid angiogram showing disappearance of the fistula after 1 year.

were excluded from the radiation field. The dose planning used multiple isocenters to improve the conformity of the target volumes that ranged from 824 to 1755 mm<sup>3</sup>. The marginal dose ranged from 13 to 15 Gy, and the maximum dose ranged from 26 to 30 Gy.

## Case Presentations

**Case 1:** A 72-year-old woman was admitted to our hospital in September 1999 with left side chemosis, progressively increasing over a 1-month period. Cerebral angiography revealed a dural AVF of the



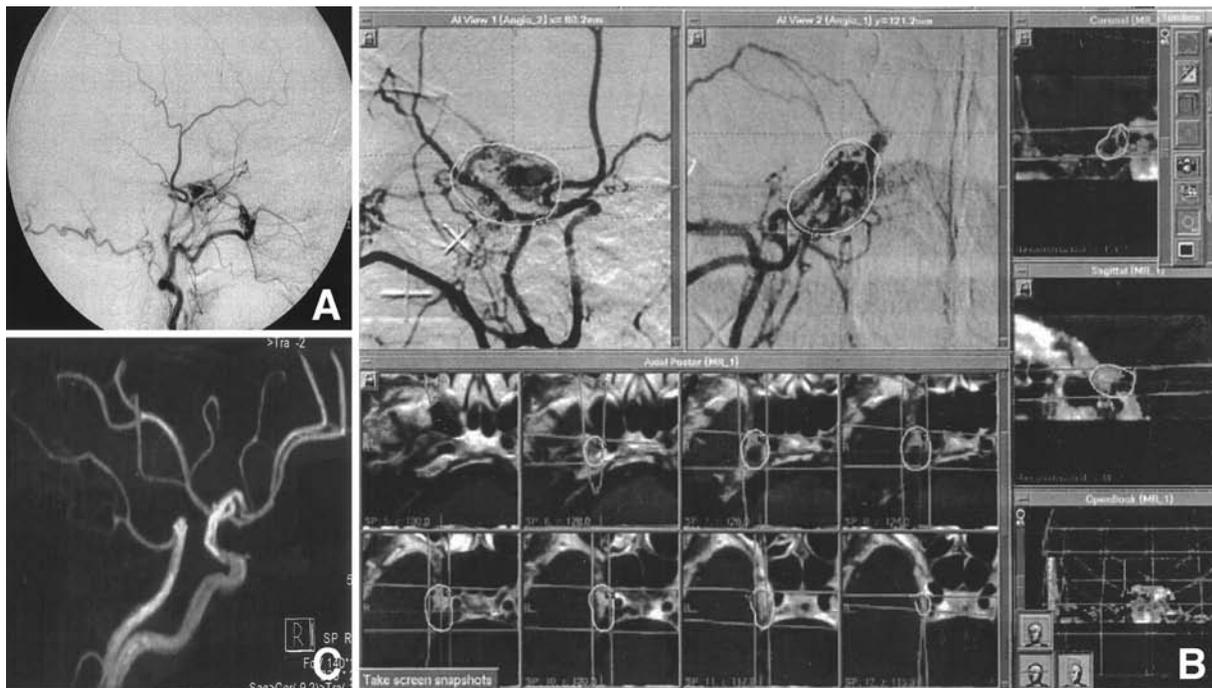
**Fig. 3** Case 3. **A:** Right carotid angiogram showing bilateral cavernous dural arteriovenous fistulas supplied by the bilateral external and internal carotid arteries and drained by the cortical veins classified as type D. **B:** Digital subtraction angiograms superimposed on magnetic resonance (MR) images showing the dose planning for radiosurgery. **C:** Follow-up MR angiogram showing that the fistulas had disappeared after 3 months.

cavernous sinus with retrograde drainage into the inferior petrosal vein. The dural AVF was classified as type D. Transvenous embolization via the inferior petrosal sinus was unsuccessful due to the tortuous route. Transarterial embolization for the meningeal branches of the ECA with polyvinyl alcohol particles was attempted, but only induced partial occlusion. Postembolization cerebral angiography revealed a dural AVF of the cavernous sinus, classified as type D converted to type B after the ECA meningeal branches were treated (Fig. 1A). Gamma knife surgery was performed with a marginal dose of 15 Gy and a maximum dose of 30 Gy. The target point was the shunt supplied from the meningohipophyseal trunk of the bilateral internal carotid arteries. Axial, coronal, and sagittal MR images with contrast medium were utilized for dose planning (Fig. 1B). Follow-up MR angiography revealed that the fistula remained at 6 months (Fig. 1C), but had disappeared at 2 years (Fig. 1D).

**Case 2:** A 71-year-old woman suffered from pulsatile tinnitus 9 months before admission to our hospital in December 2000. Cerebral arteriography revealed a cavernous dural AVF classified as type D (Fig. 2A). Stereotactic gamma knife surgery was performed

with a marginal dose of 15 Gy and a maximum dose of 30 Gy with a volume of 891.9 mm<sup>3</sup> (Fig. 2B). Follow-up MR angiography revealed that the fistula had disappeared at one year (Fig. 2C).

**Case 3:** A 79-year-old woman was admitted to our hospital in November 2001 with bilateral ptosis and chemosis persisting for 2 months. Physical examination revealed left abducens nerve paresis, ocular hypertension, and slowly progressive dementia. Cerebral arteriography showed bilateral cavernous dural AVFs classified as type D. The dural AVFs displayed bilateral arterial supply from both ECAs and internal carotid arteries with venous cortical drainage (Fig. 3A). MR imaging showed edema in the temporal lobe suggesting venous congestion, with the risk of venous ischemia leading to progressive dementia. Stereotactic gamma knife surgery was performed with a marginal dose of 13 Gy and a maximum dose of 26 Gy with a volume of 1755.3 mm<sup>3</sup> (right 1000 mm<sup>3</sup>, left 755.3 mm<sup>3</sup>) (Fig. 3B). Manual carotid artery compression was performed after the radiosurgery due to increasing intraocular pressure. Follow-up MR angiography revealed that the fistulas had disappeared after 3 months (Fig. 3C).



**Fig. 4 Case 4. A: Right carotid angiogram showing a dural arteriovenous fistula supplied by the right external and internal carotid arteries classified as type D. B: Digital subtraction angiograms superimposed on magnetic resonance (MR) images showing the dose planning for radiosurgery. C: Follow-up MR angiogram showing that the fistula had disappeared after 3 months.**

**Table 1 Summary of the cases**

Case No.	Age/Sex	Type*	Symptoms and signs	Marginal dose (Gy)	Maximum dose (Gy)	Total volume (mm <sup>3</sup> )	Follow up (months)
1	72/F	D→B	chemosis	15	30	824	32
2	71/F	D	pulsatile tinnitus	15	30	891.9	24
3	79/F	D	ptosis, chemosis, abducens nerve paresis, dementia	13	26	1755.3 (rt 1000, lt 755.3)	14
4	67/F	D	exophthalmus, chemosis	15	30	1500	26

\*According to Barrow's classification.<sup>2)</sup>

**Case 4:** A 67-year-old woman was admitted to our hospital in May 2000 with right exophthalmos and chemosis persisting for 5 months. Ocular examination revealed ocular hypertension and tortuous dilated retinal vessels. Angiography showed the dural AVF was supplied by the right ECA and internal carotid artery (Fig. 4A). Stereotactic gamma knife surgery was performed with a marginal dose of 15 Gy and a maximum dose of 30 Gy with a volume of 1500 mm<sup>3</sup> (Fig. 4B). Follow-up MR angiography revealed that the fistula had disappeared after 3 months (Fig. 4C).

## Results

All patients experienced symptomatic relief and improvement of symptoms beginning at 1 to 3 months after treatment. The follow-up period ranged between 14 and 32 months (mean 24 months). The clinical characteristics and results are shown in Table 1. Symptoms of chemosis or tinnitus improved within one week and resolved within 2 months. Intraocular pressures decreased by one month in Cases 1 and 3. Ocular symptoms and hypertension improved within 3 months in Case 4. MR angiography and DSA demonstrated total

obliteration of the dural CCF within 3 months except in Case 1. None of the patients experienced recurrence of symptoms before the disappearance of the AVF.

### Discussion

Our preliminary results suggest that stereotactic radiosurgery is an effective treatment for low-flow dural CCF with multiple shunts (Barrow type D). However, the radiosurgical indications for such lesions remain unclear, and the treatment strategy should consider the possibility of endovascular treatment, CCF flow, patient age, and informed consent. Elderly patients have various backgrounds of medical conditions and social factors, which may require less-invasive treatments. As in Case 3, venous ischemia due to dural AVF can cause progressive dementia,<sup>3)</sup> and such patients require extensive nursing care with constant observation. Therefore, radiosurgery is preferable to other treatments for post-treatment management.

Unsuccessful embolization of the dural AVF after several attempts is another indication for gamma knife surgery.<sup>9,12)</sup> Clinical improvement can be achieved in some cases of Barrow type D CCF that convert to type B after endovascular treatment, but for how long such cases should be followed up is unclear.<sup>10)</sup> As in Case 1, transvenous embolization via the inferior petrous sinus was unsuccessful and transarterial embolization did not achieve the desired results after 2 months. Therefore, gamma knife surgery is an alternative to conservative treatment.

Low-flow dural CCF is a third indication for radiosurgery. Low-flow dural CCFs have multiple shunts supplied mainly from the lateral wall and inferior wall from the meningeal branches of the ECA. The target should include these ECA feeders for fistulas of type D. The hemodynamic changes caused by only partial ECA occlusion are sufficient to lead to spontaneous resolution of the entire fistula. Aggressive signs such as cortical vein drainage and increased intraocular pressure indicate embolization is required to alleviate the symptoms and to prevent further complications of the arteriovenous shunting.<sup>4)</sup> Progressive visual loss results from a combination of reduced arterial perfusion, orbital venous hypertension, and accompanying glaucoma. Stereotactic radiosurgery will obliterate the dural AVF and prevent deterioration of visual acuity, but the effect might be slower than by endovascular treatment. Therefore, carotid compression can be performed subsequent to radiosurgery to help promote progressive thrombosis, so careful observation

of the patient's condition is required after treatment.

Follow up of the patients must include special regard for radiation injuries. The side effects of radiosurgery for dural fistulas are unknown, so we used the lowest effective doses. We ensured that the optic nerves or brainstem received no more than 8–10 Gy of radiation.<sup>1,5,8,13)</sup>

Recent advances of MR angiography have provided less-invasive methods for the identification of AVFs, but cavernous sinus and inferior petrosal sinus flow signals may cause false-positive findings in patients without symptoms or signs of CCF.<sup>3)</sup> However, MR angiography is useful for following up patients treated for dural CCF.<sup>10)</sup>

Stereotactic radiosurgery should be considered as an effective treatment alternative for elderly patients with low-flow types of CCF, or if endovascular treatment has failed.

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### Commentary

The authors have reported the successful utilization of stereotactic radiosurgery in the treatment of four patients with spontaneous dural carotid cavernous fistulas. The primary advantage of this therapeutic modality is the fact that it is minimally invasive. The disadvantages are the longer interval between treatment and desired therapeutic effect, as well as the long-term risks of radiation injury. The outcome in these four cases was universally excellent and this report underscores the importance of adding stereotactic radiosurgery to the therapeutic armamentarium for treating spontaneous dural carotid cavernous fistulas. Considering the low risk and high success associated with endovascular techniques, I believe the indications for stereotactic radiosurgical treatment of these lesions will be sparse.

I would offer two cautionary notes. One should be extremely cautious in relying upon MR angiography to document complete obliteration of these lesions. Despite advances in this technology, false-negative studies are not uncommon and catheter angiography remains the gold standard. Secondly, the use of stereotactic radiosurgery for a patient with a spontaneous dural fistula associated with leptomeningeal venous drainage (Borden type II or III) may expose

the patient to unnecessary risk. These lesions (such as the fistula in Case 3 in this series) are associated with an extremely aggressive natural history with high risk of intracerebral hemorrhage and death.<sup>1)</sup> Even a two to three month delay in therapeutic benefit may place these patients at unnecessary risk given the high likelihood of success with more rapid endovascular therapeutic options. The overwhelming majority of these lesions can be treated through transvenous approaches and those rare lesions that do not lend themselves to endovascular transvenous access can be safely and effectively treated by direct transorbital puncture of the cavernous sinus through the superior orbital fissure.<sup>2,3)</sup>

Despite these minor criticisms, the authors are to be congratulated for documenting these four cases with excellent outcomes and reminding us of the potential role for stereotactic radiosurgery in the treatment of selected lesions.

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The authors reported their experiences of stereotactic radiosurgery in four patients with spontaneous dural carotid-cavernous fistulas (CCFs). They concluded that stereotactic radiosurgery was a useful method of treating CCFs in elderly patients, cases involving low-flow CCFs, and cases in which endovascular treatment had failed. As the radiation target included not only the dura, which contained arteriovenous fistulas, but also the venous compartment of the cavernous sinus, it may need to be ascertained whether or not the treatment had a detrimental effect on the venous sinus (obliteration). If this were the case, the radiation might convert a benign fistula involving the venous drainage into a dangerous (malignant) one. Although the authors had to use low dose radiation to avoid injury to the cranial nerves in the cavernous sinus, it

would be preferable to define a rationale of low dose radiation for effective treatment of particular CCFs. In spite of these issues, the authors are to be commended for their interesting study, which demonstrated that stereotactic radiosurgery remains an effective treatment alternative for selected cases of dural CCFs.

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This article shows the possible positive effect of stereotactic gamma knife surgery for spontaneous dural carotid-cavernous fistulas. Although there have been several reports about radiation therapy, including gamma knife surgery, which type of dural fistula is suitable for stereotactic radiosurgery and how the target field is decided are still controversial. This article might provide some new accuracy for the application of stereotactic radiosurgery to dural fistulas.

Although gamma knife surgery is said to be less invasive than surgery and endovascular treatment, some recent reports suggest long-term side effects of gamma knife surgery. Those late effects should be closely followed in the present cases. In treating spontaneous dural carotid-cavernous fistulas by endovascular treatment, transvenous obliteration of the cavernous sinus with or without transarterial embolization is usually indicated. When a patient's eyesight is rapidly worsening or cortical venous reflux is observed, immediate effective treatment is required. In such cases, endovascular treatment is indicated rather than radiation therapy. However, there are some cases in which complete cure will not be achieved by endovascular treatment, so stereotactic gamma knife surgery should then be considered.

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