External Manual Carotid Compression is Effective in Patients with Cavernous Sinus Dural Arteriovenous Fistulae

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Summary

External manual carotid compression is a non-invasive method to treat patients with cavernous sinus dural arteriovenous fistulae (CS-DAVF). We studied a group of patients with CS-DAVF to identify factors that made cure by compression therapy possible.

We treated 23 patients with CS-DAVF without cortical venous drainage or a recent decline in visual acuity by compression therapy. All were followed up by magnetic resonance angiography (MRA) at one, three, six, and 12 months after treatment and the characteristics of the imaging findings, their neurological symptoms, and the patterns of symptom improvement were examined.

In group A ($n=8$), cure was achieved by manual carotid compression; in the other 15 patients (group B), cure was not obtained. Group B manifested significantly higher ocular pressure and a significantly longer interval between symptom onset and treatment by manual carotid compression. In group A, venous drainage was via the superior orbital vein (SOV) with/without involvement of the inferior petrosal sinus (IPS); closure of the CS-DAVF occurred within 4.1 months after the start of treatment. In three patients symptom improvement progressed steadily and gradually. The other five cured patients experienced transient worsening of their symptoms at two to four months after the start of

treatment, these resolved within four to seven months

Manual carotid compression was effective in patients without retrograde venous CS-DAVF drainage or a severe decline in visual acuity. The factors that rendered cure by compression therapy possible were lower ocular pressure and a shorter interval between symptom onset and the start of treatment. Venous drainage in those patients was exclusively via the SOV without involvement of the IPS.

Introduction

In the presence of cavernous sinus dural arteriovenous fistulae (CS-DAVF), dural branches of the internal and/or external carotid arteries communicate with the cavernous sinus (CS). Clinical symptoms include conjunctival chemosis, proptosis, extraocular nerve palsy, retro-orbital pain, and subjective/objective bruit^{1,2}. While the prognosis of CS-DAVF is generally favorable³, some patients with retrogradely-draining CS-DAVF presented with brain swelling or subcortical hematoma^{4,5,6}.

CS-DAVF have been treated by transarterial or transvenous embolization⁷, surgery⁸, and radiosurgery⁹. Since some of these procedures carry a potential risk for severe complications, treatment decisions must consider individual criteria. External manual carotid compression, a non-invasive, low-cost form of treatment, is

Figure 1 Clinical course of symptom abatement in group A patients. Patients 4-8 experienced transient symptom exacerbation before achieving cure by external manual carotid compression.

often used in patients with CS-DAVF¹⁰. However, factors that contribute to its success and the closure and healing of these lesions remain unclear. Therefore, we studied the imaging findings, neurological symptoms, and improvement patterns of CS-DAVF patients cured by manual carotid compression.

Patients and Method

Between 1996 and 2004, we treated 48 patients (35 women and 13 men aged from 48-76 years) with CS-DAVF at our department. Baseline angiograms were obtained for all 48 patients to document the size and location of their lesions, and the type of venous drainage of the fistulae. All patients also underwent ophthalmologic examination including visual acuity, visual field, and ocular pressure by a neuroophthalmologist.

Of the 48 patients, 23 highly compliant and motivated patients without cortical venous drainage or a recent decline in visual acuity

were treated with manual carotid compression therapy. The patients included in this study provided prior informed consent and the investigation was approved by the Ethics Committee of Kumamoto University.

Manual carotid compression

The patients were instructed to compress the carotid artery and jugular vein with the contralateral hand for ten seconds several times each hour (about six to 15 times per day). Then, using gradually increasing pressure, they compressed the carotid artery and jugular vein until pulsation was no longer palpable. We required use of the hand contralateral to the lesion site so that if weakness developed unbeknownst to the patient, the compressing hand would fall and treatment automatically stop. The patients performed the external self-treatment while sitting or lying down; the initial sessions were monitored by a physician to observe signs of neurological symptoms and bradycardia. If the initial 10-sec compression treatments

were tolerated without eliciting symptoms, their duration was gradually increased to a maximum of several minutes per compression. Patients who reported audible orbital bruit were instructed to continue the compression until the bruit ceased.

Post-treatment, all 23 patients were followed up with magnetic resonance angiography (MRA) studies at one, three, six and 12 months. Clinical cure was defined as complete resolution of the signs and symptoms attributable to the fistula. Patients were judged as cured if their symptoms disappeared, no new symptoms developed, and follow-up imaging showed disappearance of the CS-DAVF. They were judged as improved if their symptoms improved significantly and MRA showed a decrease in the size of the CS-DAVF.

We used the Mann-Whitney U-test to determine the statistical significance of differences. Statistical significance was defined as $p < 0.05$.

Results

Of 23 patients treated by external manual carotid compression, seven achieved complete clinical cure and one improved (Group A). The other 15 patients were not cured (Group B); their symptoms progressed or failed to improve in the course of six months. These patients underwent transvenous- ($n=10$), transarterial- ($n=2$), or combined transarterial-transvenous embolization ($n=3$). The interventions produced cure in 13 patients; the remaining two patients showed improvement.

All eight group A patients had chemosis, six manifested diplopia from ophthalmoplegia and one each had proptosis and diminished visual acuity. Their ocular pressure ranged from 16-28 mmHg (mean 22.5 mmHg). The interval between symptom onset and the inception of manual carotid compression ranged from 1.5-5.5 months (mean 3.5 months). Of the 15 group B patients, 13 (86.7%) had chemosis, 11 (73.3%) had diplopia from ophthalmoplegia, five (33.3%) had proptosis, and four (26.7%) manifested diminished visual acuity. Their ocular pressure ranged from 18-32 mmHg (mean 26.7 mmHg) and the interval between symptom onset and the start of manual carotid compression ranged from 3.5-7.5 months (mean 5.8 months). The two groups did not differ with respect to their clinical symptoms, however, in group B the mean ocular pressure was signifi-

Figure 2 A) Right external carotid angiogram, anterior-posterior view, showing shunt flow into the right cavernous sinus. B) Right internal carotid angiogram, lateral view, showing a dural arteriovenous fistula at the posterior cavernous sinus. Drainage is toward the right inferior petrosal sinus.

cantly higher ($p < 0.02$) and the interval between onset and treatment longer ($p < 0.03$), than in group A (table 1).

Angiographically, the CS-DAVF in group A was on the right side in two, the left side in three, and bilateral in three patients; in group B, four lesions were on the right, and five on the left side, six were bilateral. We categorized the feeding arteries according to the classification of Barrow et Al¹¹. Type B fistulae were fed only by the external carotid artery, type C by

Figure 3 A) As pre-treatment MRA demonstrated hyperintensity in the right cavernous sinus, we suspected abnormal shunt flow via a dural arteriovenous fistula. B) MRA obtained six months after the start of manual carotid compression treatment shows complete disappearance of the shunt flow.

only the dural branches of the internal carotid artery, and type D by both the external and internal carotid artery. As shown in table 1, there was no difference between the two groups with respect to the laterality of the lesions or their feeders.

In our study population, drainage was into the superior orbital vein (SOV), the superior petrosal sinus (SPS), the inferior petrosal sinus (IPS), and the contralateral CS through the intercavernous sinus; none of the patients manifested retrograde venous drainage. In group A, drainage involved only the SOV ($n=6$), the SOV plus the IPS ($n=2$), or the SOV plus the contralateral cavernous sinus ($n=1$). In group B, 12 of the 15 fistulae drained into the SOV; in six cases the IPS was involved (table 1). While there was no significant difference in the drainage route between the two groups, the SOV was the sole drainer of significantly more group A than group B fistulae ($p<0.02$).

The number of compressions applied per day ranged from seven to 13 times (mean 8.7 times) in group A and from six to 15 times (mean 9.1 times) in group B. The difference in the number of compressions applied by each group was not significant. In cured patients (group A), closure of the fistula occurred at 2.5 to seven months (mean 4.1 months) after the start of manual carotid compression therapy. In three patients improvement was steady and gradual and re-

quired compression therapy ranging from 0.5-2.5 months (mean 1.7 months). The other five patients experienced transient symptom exacerbation before achieving cure and therapy was required for a mean of 5.6 months (range four to seven months). The transiently worsening symptom of visual disturbance exacerbation manifested at two to four months (mean 3.2 months) after the start of therapy. Continued compression therapy for two to three months (mean 2.5 months) led to their resolution (figure 1).

Illustrative cases

Case 3

This 63-year-old woman had a two month history of double vision. Cerebral angiography demonstrated a right CS-DAVF fed mainly by multiple small meningeal branches of the right external and internal carotid arteries; drainage was via the right IPS (figure 2). She was instructed to perform intermittent manual compression of the right cervical carotid artery. MRA performed three months after the start of therapy demonstrated shrinkage of the CS-DAVF; her double vision was improved. She continued compression therapy and an MR angiogram acquired six months later showed complete closure of the right CS-DAVF (figure 3). Opacification of the right CS-DAVF and her double vision disappeared. Her post-treatment course

Figure 4 A) Left external carotid angiogram, anterior-posterior view, showing shunt flow into the left cavernous sinus. Drainage is toward the left inferior petrosal sinus (IPS) and the contralateral IPS through the inter-cavernous sinus. B) Left internal carotid angiogram, lateral view, showing a dural arteriovenous fistula at the posterior cavernous sinus. Drainage is toward the left IPS and left superior ophthalmic vein (SOV).

was uneventful. MRA study 12 months after treatment revealed no recurrence of CS-DAVF.

Case 6

On admission, this 62-year-old woman suffered from left orbital pain that had suddenly begun three months earlier. Chemosis was absent and she had no visual disturbance. A cerebral angiogram demonstrated a left CS-DAVF fed mainly by multiple small meningeal branches of the bilateral external and internal carotid arteries; drainage was via the bilateral IPS and left SOV (figure 4). She was instructed to perform intermittent manual compression of the left cervical carotid artery. At three months after the start of therapy she reported gradual improvement of her orbital pain, however, MRA failed to show disappearance of the lesion (figure 5) and she developed proptosis and orbital congestion. Angiography demonstrated obstruction of the left IPS and stasis of contrast medium in the left SOV. These findings indicated the development of thrombosis in the left CS (figure 6) and she was instructed to continue manual carotid compression. After three months, she manifested a striking improvement of her orbital findings. MRA performed six months later showed complete closure of her left CS-DAVF (figure 7). Opacification of the left CS-DAVF and her double vision disappeared. Her post-treatment course was un-

eventful. MRA study 24 months after treatment revealed no reappearance of CS-DAVF.

Discussion

Although the mechanism(s) of spontaneous CS-DAVF closure were unclear, Bennett et al¹² reported a CS-DAVF that became asymptomatic following a transfemoral angiogram. Newton and Hoyt¹³ noted spontaneous closure of a CS-DAVF in five patients; in three the CS-DAVF abated clinically following carotid angiography. In these cases, the disappearance of the fistulae was thought to be precipitated by post-angiography manual carotid compression. External manual carotid compression, a maneuver that facilitates thrombus formation in the CS, is an accepted treatment for CS-DAVF except in cases with cortical venous drainage and progressive visual decline. Halbach et al³ reported that among 30 of their patients treated by carotid jugular compression, seven did and 23 did not achieve complete cure. Higashida et al¹⁰ found that 15 out of 71 patients subjected to this treatment manifested complete fistula closure; none of these 15 patients exhibited clinical or angiographic signs of recurrence at one year post-treatment. In their series, closure occurred from several days to six months after the start of compression therapy. Based on their and our observations, we recommend

Figure 5 A) Pre-treatment MRA demonstrating hyperintensity in the left cavernous- and inter-cavernous sinus suspicious of abnormal shunt flow of a dural arteriovenous fistula. B) MRA obtained three months after the start of compression treatment shows resolution of the shunt flow. C) MRA obtained six months after the start of treatment shows complete disappearance of the shunt flow.

external manual carotid compression in CS-DAVF patients without rapidly progressive visual deterioration, neurological symptoms, or other complicating factors. The results of careful serial follow-up and angiographic studies help to determine whether other, more definitive therapy is necessary.

Our search of the literature found few reports describing the characteristics of CS-DAVF curable by manual carotid compression. Based on their clinical features and imaging

Figure 6 Late-phase left internal carotid angiogram, lateral view, showing no flow in the left IPS. Note residual medium in the left SOV. The flow is very slow.

characteristics, Apsimon et al¹⁴ divided 34 CS-DAVF patients into four groups. Their seven patients with anterior cavernous fistulae draining primarily into the SOV were the most amenable to manual carotid compression as the sole treatment; cure was achieved in four of these patients. In our series, successfully-treated CS-DAVF patients (group A) also exhibited characteristic features; the SOV was recognized angiographically as the only venous drainage route, and compared to group B, their mean ocular pressure was significantly lower and the interval between symptom onset and the start of manual compression treatment was significantly shorter. Moreover, among our eight patients cured by manual compression, five experienced a transient decline in visual acuity; in four of these, venous drainage involved the SOV and IPS. We posit that in these patients, the IPS was thrombosed before occlusion of the SOV so that this vessel was temporarily the only venous drainage route before complete thrombosis of the shunting point of the CS. Under these conditions, orbital pressure was elevated but declined gradually after complete thrombosis of the CS. Patients who experience exacerbation of visual symptoms after the start of manual compression therapy must be monitored closely and this therapy should only be continued in the absence of prolonged progressive worsening. Our patients with transient symptom exacerbation during compression therapy achieved complete cure during the subsequent two to three month period.

Table 1 Summary of 23 patients with cavernous sinus dural arteriovenous fistulae who underwent external manual carotid compression therapy.

n	Group A 8	Group B 15	
Symptoms			
chemosis	8 (100%)	13 (86.7)	n.s.
diplopia	6 (75)	11 (73.3)	n.s.
proptosis	1 (12.5)	5 (33.3)	n.s.
visual disturbance	1 (12.5)	4 (26.7)	n.s.
ocular pressure (mmHg)	16-28	18-32	
mean ocular pressure (mmHg)	22.5	26.7	0.02
duration between onset and treatment (months)	1.5-5.5	3.5-7.5	
mean (months)	3.5	5.8	0.03
Angiographical findings			
site			
right	2 (25.0)	4 (26.7)	
left	3 (37.5)	5 (33.3)	n.s.
bilateral	3 (37.5)	6 (40.0)	
artery			
Barrow Type B	2 (25.0)	3 (20.0)	
Barrow Type C	1 (12.5)	2 (13.3)	n.s.
Barrow Type D	5 (62.5)	10 (66.7)	
Drainer			
SOV	8 (100)	12 (80.0)	n.s.
SPS	0	2 (13.3)	n.s.
IPS	2(25.0)	6 (40.0)	n.s.
con-CS	1 (12.5)	3 (20.0)	n.s.
multiple draining system	3 (37.5)	8 (53.3)	n.s.
SOV only	6 (75)	4 (26.7)	0.02
N.S.: not significant, SOV: superior orbital vein, SPS: superior petrosal sinus, IPS: inferior petrosal sinus, CS: cavernous sinus			

Miki et Al¹⁵ encountered three patients with CS-DAVF who suffered retinal hemorrhage, probably due to central retinal vein occlusion, during manual carotid compression treatment. Komiyama et Al¹⁶ reported transient monocular blindness during manual carotid compression in two patients with DAVF; all of these patients experienced darkness in the visual field immediately after manual carotid compression.

According to Seeger et Al², external compression of the cervical carotid artery and the jugular vein acts synergistically to produce thrombosis of the fistula. Compression of the cervical carotid artery induces a transient collapse in the CS-DAVF, thereby altering the hemodynamic flow to both the CS tributaries and within the CS per se. Simultaneously, compression of the jugular vein decreases venous outflow,

raising the pressure within the CS. This may temporarily equalize the pressure between the carotid artery inflow to the fistula and the outflow from the CS and result in stasis and thrombosis within the CS or its smaller tributaries. In several of our patients, iterative MRA studies made it possible to follow the progressive development of thrombosis elicited by repeated compression of the carotid artery and jugular vein. As manual carotid compression can induce thrombus formation in the CS and occlusion in the central retinal vein due to intentionally-reduced retinal perfusion pressure, it also reduces the perfusion pressure in the retina and anterior optic nerve, resulting in a transient visual decline¹⁶.

In some patients, carotid compression therapy may stimulate the vagus nerve located in the carotid sheath and thereby elicit vasovagal reactions resulting in cardiac arrhythmias including

sinus bradycardia, atrioventricular block, systole, and syncope. In particular, elderly patients with significant atherosclerotic disease are at increased risk for neurological deficits and stroke from compromised carotid blood flow and dislodged emboli³. None of our 23 patients suffered complications attributable to manual carotid compression, possibly because we used this treatment modality only in CS-DAVF patients without retrograde venous CS-DAVF drainage or a severe decline in visual acuity.

Conclusions

The pertinent factors in CS-DAVF patients who achieved cure by external manual carotid compression therapy were a lower ocular pressure, a shorter interval between symptom onset and treatment, and venous drainage solely via the SOV without involvement of the IPS.

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