

## SHORT REPORT

## Intracranial dural arteriovenous fistulas with or without cerebral sinus thrombosis: analysis of 69 patients

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**Objectives:** To compare the characteristics of dural arteriovenous fistulas (AVFs) with or without cerebral sinus thrombosis (CST), and to analyse the determinants of aggressive manifestations in patients with dural AVF.

**Methods:** We investigated 69 patients aged 51.4 (SD 15) years who were diagnosed as having dural AVF. According to the location of the lesion and venous drainage pattern, dural AVF was classified into three sites (cavernous sinus, large sinus, and other) and five types (by Cognard's method). Aggressive manifestations of dural AVF were defined as intracranial haemorrhage, venous infarction, seizure, altered mental status, and intracranial hypertension. The diagnosis of CST was based on cerebral angiography. Logistic regression methods were used to analyse the determinants of aggressive manifestation in patients with dural AVF.

**Results:** CST was found in 39% of the patients with dural AVF. It was located at almost either the sinus around the dural AVF or the downstream venous flow pathways of the dural AVF. There was no significant difference with regard to sex, location, or type of dural AVF between patients with dural AVF with and without CST. The location "other sinuses" and the type of dural AVF "IIb/IIa-b/III/IV/V" were significantly related to aggressive manifestations of dural AVF (odds ratio 19 ( $p=0.001$ ) and 5.63 ( $p=0.033$ ), respectively). Presence of CST in patients with dural AVF had an odds ratio of 4.25 ( $p=0.12$ ) for development of aggressive manifestations.

**Conclusions:** CST affects two fifths of patients with dural AVF. The location and type of dural AVF are major determinants of aggressive manifestations in patients with dural AVF.

Intracranial dural arteriovenous fistulas (AVFs) are abnormal arteriovenous shunts localised to the intracranial dura mater.<sup>1-3</sup> Some mechanisms that have been proposed for the development of dural AVFs are based on the theory of the opening up of pre-existing arteriovenous communications and the angiogenesis theory, which suggests that angiogenesis is a result of intracranial venous hypertension.<sup>3-9</sup> In both theories, cerebral sinus thrombosis (CST) may play an important role. The shunt between the artery and sinus induces turbulent flow into the venous sinus that causes intimal injury leading to luminal thrombosis.<sup>10-11</sup> In addition, secondary CST resulting from dural AVF may amplify venous hypertension and induce retrograde flow in the sinus.<sup>5-6</sup>

Despite the strong relation between dural AVF and CST, no large in-depth study of the features of patients with both dural AVF and CST has been carried out as yet. The aims of the present study were to compare the characteristics of dural AVF with or without CST, to delineate the relation between the location of dural AVF and CST, and to find the determinants of aggressive manifestations of dural AVF.

## METHODS

The total of 72 consecutive patients who had been admitted to the National Taiwan University Hospital from May 1995 through August 2002 who were diagnosed as having dural AVF were initially included in the study. The National Taiwan University Hospital is a tertiary referral medical centre and about 1000 patients with acute stroke are referred each year.<sup>12</sup> The patients' records were reviewed retrospectively for details of demographic data and clinical manifestations. The diagnosis of dural AVF and CST was confirmed by cerebral catheter angiography. Three patients were excluded because of incomplete data, thus 69 patients were included in the analysis.

The location of the dural AVF was classed as cavernous sinus, large sinus, or "other" because these result in different clinical presentations.<sup>2-13</sup> The location "large sinus" included transverse, sigmoid, and superior sagittal sinuses. "Other" locations were those other than the cavernous and large sinuses. We further classified dural AVFs according to Cognard's method<sup>13</sup> into five types according to the venous drainage pattern. Based on the clinical manifestations, dural AVFs were divided into "aggressive" and "non-aggressive" groups similar to previous reports.<sup>2-13-14</sup> The aggressive group included patients with intracranial haemorrhage, venous infarction, seizure, altered mental status, and intracranial hypertension. The non-aggressive group included patients who had tinnitus, ocular symptoms not related to intracranial hypertension, and cranial neuropathy.

The angiographic characteristics of CST included in the analysis were total occlusion of sinus (such as abrupt cessation of contrast medium passage in the dural sinus); reversed flow in the non-involved part of the sinus and reflux into the other sinus or cortical vein; and stasis of the contrast medium at the point of thrombosis.

The  $\chi^2$  test was used to determine the differences between the groups for some features (including sex, presence of CST, location, and type of AVF). Fisher's exact test was used when any one of the frequencies was less than 5. Student's *t* test was used to determine the differences in ages. A logistic regression model was used to analyse the determinants of aggressive manifestation of dural AVF. The model included age, sex, the type and site of dural AVF, and the presence of CST. A *p* value of less than 0.05 was considered significant. SPSS statistical software (version 10.0, SPSS) was used for the statistical analyses.

## RESULTS

Of the 69 patients, 26 were men and 43 women (mean age (SD) at diagnosis 51.4 (14.7) years, range 23–81). Twenty four patients (35%) had aggressive symptoms. There were no sex differences with regard to the presence of CST ( $p=0.54$ ).

**Abbreviations:** AVF, arteriovenous fistula; CI, confidence interval; CST, cerebral sinus thrombosis; OR, odds ratio

**Table 1** Patients with dural arteriovenous fistulas with and without aggressive manifestations: results of univariate analyses\*

	Dural arteriovenous fistulas		p value
	With aggressive manifestations (n = 24)	Without aggressive manifestations (n = 45)	
Age in years (SD)	47 (15)	54 (14)	0.054
Men (%)	15 (63)	11 (24)	0.002
Presence of cerebral sinus thrombosis (%)	10 (42)	17 (38)	0.75
Type of dural AVF (%)			
I and IIa	6 (25)	36 (80)	<0.001
IIb, IIa+b, III, IV, and V	18 (75)	9 (20)	
Location of dural AVF (%)			
Cavernous sinus	0 (0)	21 (47)	<0.001
Transverse/sigmoid/superior sagittal sinuses	5 (21)	17 (38)	
Other sinuses†	19 (79)	7 (16)	

\*Values are number (percentage) except age.

†Other sinuses include all locations other than the cavernous sinus and large sinus (transverse, sigmoid, and superior sagittal sinuses).

The most frequent location of dural AVF was the cavernous sinus (21 (30%)), followed by the skull base (13 (19%)), transverse sinus (12 (17%)), dural convexity (8 (12%)), sigmoid sinus (7 (10%)), torcular Herophili (5 (7%)), and superior sagittal sinus (3 (4%)). All 21 patients with cavernous sinus dural AVF developed symptoms of ophthalmopathy—that is, chemosis, ocular palsy, and exophthalmoses. Eighteen (82%) of 22 patients with large sinus dural AVF had pulsatile tinnitus. No patient with cavernous sinus dural AVF presented with aggressive symptoms, compared with 5 (23%) and 19 (73%) patients in the groups of large sinus and “other” (n = 26), respectively who had aggressive symptoms. Despite the prominent correlation between the characteristics of dural AVF and its location, there were no significant differences in the presence of CST between the different locations of dural AVF (p = 0.2).

Type I (n = 24) and IIa (n = 18) AVFs were the commonest, but yielded the lowest frequencies of aggressive manifestations (17% and 11%, respectively). Those with venous drainage including the cortical vein (type IIb (n = 3), IIa+b (n = 9) and III (n = 10)) had higher frequencies of the aggressive presentations (67%, 44%, and 70%, respectively). All of the patients with type IV (n = 3) and V (n = 2) dural AVF presented with aggressive symptoms. Nevertheless, there were no significant differences in the presence of CST among different types of dural AVF (p = 0.82).

CST was diagnosed in 27 (39%) patients with dural AVF. Among these, the commonest locations of CST were the transverse sinus (9 (33%)) and cavernous sinus (9 (33%)), followed by the sigmoid sinus (7 (26%)) and superior sagittal sinus (5 (19%)). Ten (37%) patients had CST at more than one location.

A relationship was found between the location of dural AVF and the site of CST. Of 11 patients with cavernous sinus dural AVF and CST, eight had thrombosis in the cavernous sinus and three had thrombosis in the venous flow downstream from the cavernous sinus. In nine patients with large sinus dural AVF and CST, the CST was just at the site of the dural AVF or near it. The CST was located at almost either the sinus around the dural AVF or the downstream venous flow pathways of the dural AVF.

Table 1 shows the features of dural AVF in patients with and without clinical aggressive manifestations. There were

**Table 2** Aggressive manifestations in patients with dural arteriovenous fistulas: results of multivariate analyses

	OR	95% CI	p value
Age in years	1.03	0.98 to 1.08	0.28
Male sex	4.67	0.98 to 22.4	0.054
Presence of cerebral sinus thrombosis	4.25	0.68 to 26.64	0.12
Type of dural AVF at type IIb, IIa+b, III, IV, and V	5.63	1.15 to 27.57	0.033
Location of dural AVF at other sinuses†	19.00	3.29 to 109.83	0.001

†Other sinuses include all locations other than the cavernous sinus and large sinus (transverse, sigmoid, and superior sagittal sinuses).  
OR, odds ratio; CI, confidence interval.

significant differences between the groups for sex (p = 0.002), and location (p < 0.001) and type (p < 0.001) of dural AVF. By the logistic regression model, only the location of dural AVF (other sinuses) (odds ratio (OR) 19, 95% confidence interval (CI) 3.29 to 109.83, p = 0.001) and the type of dural AVF (type IIb/IIa+b/III/IV/V) (OR 5.63, 95% CI 1.15 to 27.57, p = 0.033) were significant determinants of aggressive manifestation of dural AVF (table 2).

## DISCUSSION

Of our patients with dural AVF, 39% also had CST. It was not uncommon to find that dural AVF and CST developed together. On analysing the features of patients with dural AVF with the presence or absence of CST, there were no significant differences between the two groups with regard to sex, or location or type of dural AVF. Neither sex nor the location or type of dural AVF was more prone to concomitant CST.

As shown in previous reports,<sup>11–14</sup> the location, other sinuses, and type, IIb/IIa+b/III/IV/V, of dural AVF were significant determinants of aggressive manifestation of dural AVF. In addition to the symptoms of the dural AVF, patients with both dural AVF and CST may develop aggressive symptoms of CST.<sup>15–16</sup> CST may also force retrograde venous drainage through the cortical veins, predisposing patients to a more aggressive neurological course.<sup>5–17</sup> In the present study, however, patients with both dural AVF and CST did not show more aggressive manifestations than those with dural AVF alone. This may have been because of the relatively small sample size of this study or the possibly special and different role of CST in patients with both dural AVF and CST compared with patients with CST alone—for example, transverse embolisation of dural AVF may lead to iatrogenic CST, but usually does not create much problem. Further large prospective studies are necessary to delineate this issue.

Two hypotheses have been proposed for the pathogenesis of dural AVF. The first is based on the physiological arteriovenous shunts between the meningeal arterial networks and the dural venous sinuses.<sup>18</sup> An increase in sinus and venous pressure—for example, by the obstruction of venous outflow by CST, may open these channels to create dural AVFs. The second hypothesis, as shown in the rat model, suggests that venous hypertension induced by an obstruction to venous outflow may reduce cerebral perfusion and lead to ischaemia, followed by angiogenesis. The aberrant angiogenic activity of the dural blood vessels would then result in arteriovenous shunting.<sup>5</sup> In both, CST may be the primary event that caused the venous hypertension. In our patients with both dural AVF and CST, we found that most of the locations of the CST were either the sinus(es) around the dural AVF or the sinus(es) in the downstream pathways of the venous flow from the dural AVF. This finding supports both hypotheses—that there is a role of CST

in the pathogenesis of dural AVF through venous hypertension. In addition, if dural AVF generated secondary CST as a result of the turbulent flow into the venous sinus,<sup>5 6 10 11 17</sup> we could predict that the thrombotic sinus would be around the location of dural AVF—this notion is supported by the results of our study.

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