CASE REPORT

Spontaneous closure of a dural arteriovenous fistula

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SUMMARY

Spontaneous closure of a dural arteriovenous fistula (dAVF) is a rare condition and only a few cases have been reported since its first description in 1976. We report delayed and progressive spontaneous closure of a dAVF after massive intracerebral hemorrhage documented by angiographic studies before and after bleeding. To our knowledge, this is the first report to document gradual closure of a dAVF by serial angiographic studies. The mechanism of spontaneous closure of dAVFs has not been fully elucidated.

We suggest different factors for consideration from previously published data and show how each of these factors can influence the others.

BACKGROUND

Only a few cases of spontaneous closure of a dural arteriovenous fistula (dAVF) have been reported.^{1–11} Various precipitating factors have been postulated which include venous thrombosis, post-angiographic changes, and intracranial hemorrhage.^{2–6} Although

Figure 1 (A) Initial transfemoral cerebral angiography of the right external carotid artery before the intracerebral bleeding, lateral view, demonstrating early shunts to the sigmoid sinus via feeders from the left occipital artery, left ascending pharyngeal artery, and left middle meningeal artery. Note the retrograde flow from the transverse sinus to the cerebral veins. (B) Initial transfemoral cerebral angiography before the intracerebral bleeding: right internal carotid artery injection, lateral view, showing a shunt to the sinus filling through a feeder from the meningohypophyseal artery. (C) Initial transfemoral cerebral angiography before the intracerebral bleeding showing the ipsilateral sigmoid sinus is occluded.

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Figure 2 Cranial CT showing acute hemorrhage in the left temporal, occipital, and parietal lobes with mass effect.

many possible mechanisms have been discussed, the subsequent events and the timeline of spontaneous closure remain unclear. We present a case of gradual closure of a dAVF after intracranial hemorrhage documented by serial angiography.

CASE PRESENTATION

A 73-year-old woman was referred to a local hospital after a generalized epileptic seizure. MRI and transfemoral cerebral angiography showed a dAVF Cognard type IIa+b of the left transverse sinus with feeders from the left occipital artery, left pharyngeal artery, left middle meningeal artery, and branches of the meningohypophyseal artery ([figure 1A, B](#)). The ipsilateral sigmoid sinus was not filled and there was reflux into the

Figure 3 Angiography of the left external and internal carotid arteries 2 weeks after the intracerebral bleeding showing decreased flow to the shunt from the feeders of the left middle meningeal and the left ascending pharyngeal artery.

Figure 4 Angiography of the left external and internal carotid arteries 4 weeks after the intracerebral bleeding indicates a further decrease in shunt volume.

cerebral veins ([figure 1C](#)). She was conservatively treated with antiepileptic medication and was discharged 3 days after admission. Future endovascular therapy was planned.

One week later the patient was admitted to our hospital after an episode of severe headache. On neurological examination she was somnolent and her pupils were asymmetric. The CT on admission revealed a large and disseminated intracerebral hemorrhage in the left temporal, occipital, and parietal lobes ([figure 2](#)). The hematoma was surgically evacuated without adverse events. Two days later a decompressive craniectomy was performed for treatment of generalized brain edema with increased intracerebral pressure refractory to conservative therapy. During both surgical procedures no attempts were made to identify and occlude the arteriovenous shunts. The patient recovered slowly after the bleeding and was sent for rehabilitation with a hemiparesis.

INVESTIGATIONS

Two weeks after the hemorrhage when the patient was scheduled for endovascular occlusion of the dAVF, a decrease in shunt flow was noted ([figure 3](#)). The endovascular embolization was therefore postponed and another angiography was planned. Two weeks later further slowing of blood flow in the dAVF was documented angiographically ([figure 4](#)). Eight weeks after the hemorrhage, angiography revealed complete closure of the dAVF ([figure 5A, B](#)). The ipsilateral sigmoid sinus was still occluded ([figure 5C](#)).

OUTCOME AND FOLLOW-UP

At follow-up 1 year after the bleeding the patient had recovered and was independent with mild aphasia and a slight hemiparesis on the right side.

DISCUSSION

Spontaneous closure of a dAVF was first reported independently by Magidson and Weinberg and by Hansen and Sogaard in 1976.^{5,7} Since then, 21 other cases have been published ([table 1](#)).

Figure 5 (A) Final angiography of the external carotid arteries 8 weeks after the intracerebral bleeding showing complete closure of the dural arteriovenous fistula. (B) Final angiography of the internal carotid also showing complete closure of the dural arteriovenous fistula. (C) Final angiography after the closure of the dural arteriovenous fistula: the ipsilateral sigmoid sinus is still occluded.

Luciani and colleagues suggested the existence of two categories of dAVFs, post-traumatic and spontaneous, which would have a different impact on non-therapeutic regression.⁴

Based on published data, we suggest consideration of four basic mechanisms which may contribute to spontaneous dAVF closure: (1) post-angiography; (2) post-hemorrhagic; (3) post-thrombotic; and (4) post-recanalization. Of course, each of these mechanisms may influence the others and contribute to closure to varying degrees in a given individual.

dAVFs caused by traumatic brain injury appear to constitute a special group, as suggested by Luciani *et al.*⁴ It is remarkable that the majority of post-traumatic dAVFs were Cognard type I and had no retrograde venous drainage. In two post-traumatic dAVFs there were no symptoms at presentation, and in three cases there were no symptoms at occlusion. This might indicate that many post-traumatic dAVFs remain undiagnosed and their spontaneous closure is underestimated.⁴ The mechanism of spontaneous closure of post-traumatic dAVFs is unclear; it may be attributed to the healing process with the

tendency to develop scar tissue, which may occlude the rather small feeders of post-traumatic dAVFs.⁴

Spontaneous closure of dAVFs after angiography can be identified as a distinct mechanism.^{3 8} Five spontaneous closures supposed to be related to cerebral angiography have been described in the literature. In one of these cases closure occurred even during angiography,⁶ and in the other patients closure occurred a few hours or days after angiography.^{3 8} The main reason for this effect may be a thrombogenic potency of the contrast medium.^{3 9} Acute hemodynamic changes during the procedure might also be relevant. In addition to the above mentioned cases, Saito and colleagues presented a patient in whom closure of a longstanding dAVF (36 months) was observed within a month after cardiac catheter angiography.¹

Intracranial hemorrhage was suggested as another cause initiating spontaneous closure of a dAVF.¹⁰ Such dAVFs were usually more complex Cognard type II or III. It seems that high-flow fistulas (Cognard II or higher) are more likely to resolve after an

Table 1 Published data on spontaneous closure of dural arteriovenous fistulae with assumed mechanism of occlusion

Author (year of publication)	Sex/age	Symptoms at diagnosis	Symptoms at occlusion	Interval between initial presentation and occlusion	Feeder	Draining sinus or sinus involved	Cognard type	Interval between first angiography and angiography at occlusion	Sinus patency at presentation/occlusion	Mechanism of occlusion
Magidson (1976)	F/39	Tinnitus, bruit	Dizziness, headache, tinnitus and bruit disappeared	52 months	MMA, OA, TBs f. ICA	TS right	I	40 months	Patent/occluded	Sinus thrombosis
Hansen (1976)	F/23	Tinnitus, bruit, headache	Tinnitus and bruit disappeared	Approx. 24 months	OA, Bs f. VA	Torc	Ila	Approx. 15 months	Unknown	Unclear
Endo (1979)	F/76	Tinnitus, vertigo, seizure, bruit	Tinnitus and bruit disappeared	97 months	OA, MMA	TS left	Ila+b	24 months	Unknown	Unclear
Bioth (1979)	M/28	Tinnitus, bruit	Tinnitus and bruit disappeared	118 months	OA, MMA, TB f. ICA, Bs f. VA	TS right	I	116 months	Patent/occluded	Post-traumatic category* (5 months after TBI)
Chaudhary (1982)	F/56	Tinnitus, bruit	Headache	36 months	OA, MMA	SS right	I	36 months	Patent/irregular	Post-traumatic category* (8 months after TBI)
Chaudhary (1982)	M/38	Seizure	None	NA	OA, MMA	CV, SSS	III	24 months	Patent/irregular	Post-traumatic category (3 weeks after TBI)
Chaudhary (1982)	M/50	None	None	NA	OA, MMA	TS right	I	1 month	Patent/irregular	Post-traumatic category (6 months after TBI)
Olutola (1983)	M/50	Epidural hematoma, headache, vomiting, tinnitus, bruit	None	NA	MMA	SPS, PP left	I	4 months	Patent/patent	Post-intracranial hemorrhage
Kataoka (1984)	M/56	Intraventricular hematoma with acute hydrocephalus, headache, seizure	Seizures	15 months	OA, STA	TS right, SSS	Ila	15 months	TS: occluded/occluded SSS: patent/patent	Post-intracranial hemorrhage*
Landman (1984)	M/60	Tinnitus, bruit	Disappearance of tinnitus and bruit directly after angiography, hearing loss	24 months	APA	JV right	I	6 weeks	JV: patent/patent	Post-angiography
Rohr (1985)	F/68	Tinnitus	Transient ischemic attack	24 months	OA, MMA	SS left	I	24 months	Patent/occluded	Sinus thrombosis
Reul (1993)	M/24	Seizures, headache, ICB	None	—	EBs f. OtA	BV, SSS	IV	0	During angiography	Post-angiography
Luciani (2001)	M/42	Tinnitus, bruit	Tinnitus disappeared	180 months	MMA, OA, PMA	Torc	I	185 months	Patent/patent	Unclear
Luciani (2001)	F/57	Tinnitus	Tinnitus disappeared	374 months	MMA, PMA	TS right	I	240 months	Patent/patent	Unclear*
Luciani (2001)	F/28	None, proptosis due to coexistence of carotid-sinus fistula	None	NA	Tb f. ICA	SPetS left	I	12 months	Patent/patent	Post-traumatic category (6 months after TBI)
Moriya (2007)	M/60	Tinnitus	Vomiting, disorientation, amnesia	7 days	MMA, OA	TS left	I	7 days	Stenosis/occluded	Post angiography
Saito (2008)	M/60	Tinnitus, diplopia	Tinnitus disappeared	36 months	MMA, OA, APA, TBs f. ICA	TS right	I	36 months	Patent/patent	Post-angiography (cardiac angiography)
Voormolen (2009)	F/53	None (2 months after thrombosis of left TS and SS)	Headache, aphasia, hemiparesis	3 months	MMA, OA, CV	CV	III	Hours	TS and SS: occluded/occluded	Post-angiography
Voormolen (2009)	F/63	Dizziness, hearing problems	Headache	Days	MMA, AEA, DBs f. OtA	CV	IV	Days	Patent	Post-angiography
Waaren (2010)	M/51	Tinnitus, headache	Headache, tinnitus disappeared	120 months	OA, MMA, PMA	TS left	Ila	Approx. 120 months	Occluded/patent	Recanalization of sinus
Clarencon (2012)	M/45	Headache (ICB)	None	—	MMA, OA	TS left	Ila	3 months	Partially occluded/occluded	Post-intracranial hemorrhage

Continued

Table 1 Continued

Author (year of publication)	Sex/age	Symptoms at diagnosis	Symptoms at occlusion	Interval between initial presentation and occlusion	Feeder	Draining sinus or sinus involved	Cognard type	Interval between angiography and angiography at occlusion	Sinus patency at presentation/occlusion	Mechanism of occlusion
Clarencon (2012)	M/60	Headache (ICB)	None	–	MMA, OA	TS right	IIb	3 months	Occluded/occluded	Post-intracranial hemorrhage*
Clarencon (2012)	M/61	Headache, dyskinesia, gait disorder (ICB)	None	–	PMA, OA	CV	III	6 months	Patent/unknown	Post-intracranial hemorrhage*
Al-Afif (2014)	F/73	Seizures	Headache, ICB	26 days	OA, MMA, APA, TBs f. ICA	TS right	IIb	4 months	Occluded/occluded	Post-intracranial hemorrhage*

*Operation or selective embolization was unsuccessful.

AEa, anterior ethmoidal artery; APA, ascending pharyngeal artery; Bs, f. VA, branches from vertebral artery; BV, basal vein; CV, cortical veins; DBs f. OVA, ethmoidal branches from ophthalmic artery; ICB, intracerebral bleeding; IVB, intraventricular bleeding; JV, jugular vein; MMA, middle meningeal artery; NA, not available; OA, occipital artery; PMA, posterior meningeal arteries; PP, pterygoid plexus; SDH, subdural hematoma; SPeS, superior petrosal sinus; SPS, sphenoparietal sinus; SS, sigmoidal sinus; SSS, superior sagittal sinus; STA, superior temporal artery; TBI, traumatic brain injury; TBs f. ICA, tentorial branches from internal carotid artery; Torc, torcular herophili; TS, transverse sinus.

increase in local intracranial pressure secondary to hemorrhage. The time period between the initial angiography and the angiography at occlusion ranged from 3 to 6 months. The subsequent induction of thrombosis might also be of relevance in these cases.

Spontaneous formation of a thrombus in the draining sinus is another possible mechanism for the closure of dAVFs. There were at least two instances in which this occurrence constituted the primary event.^{5 11} In both cases occlusion was associated with typical symptoms of acute cerebral sinus thrombosis such as severe headache and dizziness. The causes of the thrombus were unknown.^{5 11} The time interval between angiography at diagnosis and occlusion in these two cases was 40 and 24 months. This long duration excludes the possibility of thrombus formation caused by the first angiography.

Warren and colleagues recently suggested another possible mechanism for spontaneous closure of dAVFs—namely, recanalization of the occluded draining sinus—which could prompt regression of the dAVF.²

It is remarkable that closure after hemorrhage in our case did not occur immediately but progressively. It is possible that high intracranial pressure and direct pressure of the hematoma compressed both the feeders and draining veins initially. Subsequent surgery for decompression of the hematoma most likely resulted in additional hemodynamic changes and possibly partial closure of the feeders. Subsequent angiographies might then have induced chronic thrombosis of the draining veins. Our case clearly shows that different mechanisms leading to spontaneous closure of a dAVF can be involved in an individual.

While several mechanisms may be involved in spontaneous closure of a dAVF, neither its occurrence nor the time frame are predictable. It would therefore be unjustified to adapt a 'wait and see' strategy in higher grade dAVFs.

Learning points

- ▶ Four basic mechanisms may contribute to spontaneous dural arteriovenous fistula closure: (1) post-angiography; (2) post-hemorrhagic; (3) post-thrombotic; and (4) post-recanalization.
- ▶ Each of these mechanisms may influence the others and contribute to closure to varying degrees in a given individual.
- ▶ While several mechanisms may be involved in spontaneous closure of a dAVF, neither its occurrence nor the time frame are predictable.

Contributors SA-A: acquisition, conception, design, analysis and interpretation of data, drafting of the manuscript, and final approval. MN: critical revision of the manuscript and final approval. FG: analysis and interpretation of data, final approval. JKK: interpretation of data, critical revision of the manuscript, final approval.

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