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Case study

Three-dimensional angioarchitecture and microsurgical treatment of arteriovenous fistulas at the craniocervical junction

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ABSTRACT

Digital subtraction angiography (DSA) is the gold standard for diagnosing vascular malformations; however, difficulties are associated with visualizing the angioarchitecture of arteriovenous fistulas at the craniocervical junction (CCJ AVFs) using DSA because of their complex regional neurovascular anatomy. The present study evaluated the application of 3-dimensional computer graphics (3D CG) to the surgical planning of CCJ AVFs.

Six patients with CCJ AVFs who underwent microsurgery and/or endovascular treatment were included. The results of DSA and 3D CG were compared in the last 3 patients. The visibility of important anatomical structures were evaluated using visibility grading scores. Clinical outcomes were assessed based on the rate of occlusion of AVFs, surgical complications, neurological status, and recurrence in long-term follow-ups.

The 3D CG images could combine arteries, veins, the spinal cord and dura mater in one single picture to evaluate the anatomy of CCJ AVFs. The image interpretation of vascular structures, particularly narrow arterial feeders, was significantly better using 3D CG than DSA (overall visibility scores, 97% vs 51%, $p = 0.001$). In all patients, the complete occlusion of AVFs was achieved by microsurgery except for 2 patients without surgical planning with 3D CG. Postoperatively, the neurological status of all patients improved or stabilized without the recurrence of AVFs (median, 5.4 years).

3D CG may help to improve the quality of the microsurgical procedures in complex AVFs. However, it should be used as a complementary diagnostic modality rather than the alternative of DSA because 3D CG has no hemodynamic information at this time.

1. Introduction

Arteriovenous fistulas at the craniocervical junction (CCJ AVFs) are rare vascular disorders that present with severe neurological symptoms such as subarachnoid hemorrhage (SAH) or venous congestion of the spinal cord [1–4]. Digital subtraction angiography (DSA) is the gold standard for the diagnosis of these lesions; however, the visualization of their angioarchitecture by DSA only is limited because of their complex regional anatomy [5–9]. Accurate preoperative diagnosis is challenging, including the visualization of feeding arteries, the AVF, the proximal draining vein, and the aneurysm, which are the targets of interruption in either microsurgery or endovascular treatment [6,8,10]. We developed 3-dimensional computer graphics (3D CG) in order to visualize complex neurosurgical diseases [11–13]. The purposes of the present study were to evaluate the clinical application of 3D CG to CCJ AVFs, describe microvascular anatomy of CCJ AVFs in detail, and assess clinical outcomes.

2. Methods and materials

CCJ AVFs were defined as arteriovenous shunts located at the C1 or C2 level of the cervical spine. The total population of patients included 6 patients with CCJ AVFs treated surgically between 2000 and 2015 at the University of Tokyo Hospital ($n = 4$) and Tokyo Metropolitan Neurological Hospital ($n = 2$). All diagnoses of CCJ AVFs were made by DSA. In the last 3patients, a 3D computer graphic analysis was conducted to assess the stereoscopic anatomy of the vascular system as well as its relationship with the dura mater and spinal cord. 3D CG was created based on the fusion of rotational angiography and myelographic CT or MRI by T.K., as described previously [11–13]. In order to compare the results of

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Table 1
Visibility grading score.

Score	Remarks
0	not identified
1	identified a putative object, but the examiner considered the structures insufficient for the simulation
2	clearly identified in the general outline, but not in detail
3	precisely identified

DSA and 3D CG, the visibility of important anatomical structures, including feeding arteries, the AVF, draining veins, and the aneurysm, were evaluated using visibility grading scores (Table 1) [14]. Anatomical structures were identified by the operating neurosurgeons (K.T. and N.H.) using surgical videos. Evaluations of 3D CG were performed by 3 blinded doctors (S.F., N.S., and S.N.) who were not involved in the surgery or image rendering.

Clinical outcomes were assessed based on the rate of occlusion of AVFs, surgical complications, neurological status (modified Rankin Scale [mRS] scores), and recurrence in long-term follow-ups. Statistical analyses were performed with the Mann-Whitney U method and IBM SPSS Statistics version 23 (IBM Corp., Armonk, NY, USA). A p value < 0.05 was considered to be significant.

This study protocol was approved by the Institutional Review Board of the University of Tokyo hospital. Since this was a retrospective and non-invasive study, written informed consent was not obtained. A public notice that provided information on this study was instead displayed on our hospital website.

3. Results

3.1. Clinical outcomes

Six patients with CCJ AVFs (median age = 55 years, male-to-female ratio = 2:1, Table 2) presented with venous congestive myelopathy (n = 4) or subarachnoid hemorrhage (n = 2). Of the 6 patients, 5 underwent microsurgery at our hospital, while the remaining patient underwent endovascular embolization at another hospital as the first-line treatment. Two patients without surgical planning with 3D CG required second-stage surgery because of the incomplete occlusion of AVFs: 1 underwent microsurgery and the other underwent endovascular treatment. Brain stem infarction occurred in 1 patient who received endovascular treatment because of occlusion of the spinal artery. Eventually, all patients achieved complete occlusion of the AVF. The neurological status of all patients improved or stabilized postoperatively. Among the 6 patients, 4 showed an independent status (mRS scores of 1 or 2), whereas 2 had a dependent status (mRS scores of 4). AVFs did not recur in the follow-up period (median, 5.4 years after surgery, range 1–15 years).

Table 3
Visibility scores among 3 patients and statistical findings.

Patient	n*	Visibility score		
		Full Score†	3D CG (%)	2D DSA (%)
4	4	36	35 (97)	17 (47)
5	4	36	36 (100)	15 (42)
6	6	54	51 (94)	32 (59)
Total	14	126	122 (97)	64 (51)
			p = 0.001	

Bold indicates statistical significance.
* The number of vascular structures assessed in each case.
† The number of structures assessed × 3 scores × 3 observers.

Fig. 1. Patient 3: dural AVF. A 76-year-old male presented with slowly progressive myelopathy because of venous congestion of the high cervical spinal cord. Right vertebral angiogram (VAG) showed an enlarged intradural vein at the C1 level; however, difficulties were associated with visualizing its complex anatomy in detail using 2D digital subtraction angiography (2D DSA) only. This patient required additional surgery because of incomplete occlusion of the AVF. Two microsurgical procedures revealed an arteriovenous (AV) shunt between the C1 radiculomeningeal artery and 2 lateral medullary veins. These proximal veins were occluded at the right C1 level.

Table 2
Clinical characteristics of 6 patients with arteriovenous fistulas at the craniocervical junction.

Patient	Age	Sex	Presentation	DSA	3D CG	Treatment		Complication	mRS		Follow-up
						Initial	Additional		pre	post	(years)
1	69	M	congestion	yes	no	surgery	no	no	3	1	15
2	79	M	congestion	yes	no	surgery	no	no	3	2	1.0
3	76	M	congestion	yes	no	surgery	surgery	no	4	4	6.8
4	64	F	congestion	yes	yes	surgery	no	no	4	4	6.3
5	68	M	SAH	yes	yes	surgery	no	no	3	2	4.5
6	46	F	SAH	yes	yes	endo*	surgery	infarction	3	2	3.0

3D CG = 3-dimensional computer graphics, DSA = digital subtraction angiography, endo = endovascular embolization, mRS = modified Rankin Scale, post = post-treatment, pre = pre-treatment.
* Endovascular treatment was performed at another hospital before the development of 3D CG.

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