### Case Report

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## Thrombosed giant aneurysm of the distal anterior cerebral artery treated with aneurysm resection and proximal pericallosal artery– callosomarginal artery end-to-end anastomosis: Case report and review of the literature

# Ken Matsushima, Masatou Kawashima, Kenji Suzuyama, Yukinori Takase, Tetsuro Takao, Toshio Matsushima

Department of Neurosurgery, Saga University, Saga, Japan

E-mail: Ken Matsushima - ken.matsushima@hotmail.co.jp; \*Masatou Kawashima - m996kawa@cc.saga-u.ac.jp; Kenji Suzuyama - suzu@karatsu.jrc.or.jp; Yukinori Takase - takase@cc.saga-u.ac.jp; Tetsuro Takao - montblanc1999da@yahoo.co.jp; Toshio Matsushima - matsuto@cc.saga-u.ac.jp \*Corresponding author

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#### Abstract

**Background:** Giant distal anterior cerebral artery (DACA) aneurysms are extremely rare, with only 32 cases reported in the literature. Most giant DACA aneurysms have features that make standard neck clipping difficult, and bypass surgery is sometimes required, although this surgery was performed in only three reported cases. This report presents the fourth case treated with bypass surgery.

**Case Description:** A 69-year-old female presented with an unruptured thrombosed giant DACA aneurysm. She underwent wrapping operation 7 years before, but radiological imaging revealed enlargement of the aneurysm at the left pericallosal artery (PerA)–callosomarginal artery (CMA) junction. Before operation, three different strategies were considered for bypass surgery in case the neck could not be clipped. Aneurysm resection and left proximal PerA–CMA end-to-end anastomosis were successfully performed under intraoperative digital subtraction angiography (DSA) and motor-evoked potential (MEP) monitoring.

**Conclusion:** Most DACA aneurysms are located at the PerA–CMA junction. In some cases, adequate retrograde flow to the distal PerA from the posterior or middle cerebral artery can be expected, making distal PerA reconstruction unnecessary. Moreover, when the distal PerA is cut, proximal PerA–CMA end-to-end anastomosis can be easily performed because of reduced tension in both vessels. We therefore conclude that this strategy should be utilized for treating such patients. We also presented here the effectiveness of intraoperative modalities, such as intraoperative DSA and MEP monitoring, for performing a safe operation.

**Key Words:** Bypass surgery, distal anterior cerebral artery, end-to-end anastomosis, intraoperative modalities, pericallosal artery, thrombosed giant aneurysm



#### INTRODUCTION

Distal anterior cerebral artery (DACA) aneurysms are rare, comprising approximately 5% of all intracranial aneurysms.<sup>[4,11,30]</sup> Most DACA aneurysms are small. These aneurysms are known for their tendency to bleed even when they are very small and for difficulty with surgical treatment. Giant DACA aneurysms are extremely rare. To our knowledge, 32 cases have been reported, including only 3 cases treated with bypass surgery.<sup>[3-7,9-13,15,17-21,23-27,29-32,34].</sup>

We successfully treated an elderly woman who presented with a partially thrombosed giant DACA aneurysm with aneurysm resection and proximal pericallosal artery (PerA)–callosomarginal artery (CMA) end-to-end anastomosis. We report here surgical treatments available for giant DACA aneurysms and the effectiveness of intraoperative modalities for performing a safe operation.

#### **CASE REPORT**

#### History

In August 2003, a 69-year-old female who had hypertension and an abdominal aortic aneurysm complained of dizziness. A computed tomography (CT) scan showed a 15-mm heterogeneous mass in the cerebral left frontal lobe [Figure 1a]. Neck clipping of this partially thrombosed DACA aneurysm was unsuccessful because of severe calcification. Therefore, wrapping operation was performed. After the operation, the patient showed no symptoms for 7 years.

In September 2010, magnetic resonance angiography at a local hospital revealed enlargement of the aneurysm, and the patient was admitted to our hospital. Neurological examinations did not show any abnormal findings. A CT scan [Figure 1b] and magnetic resonance imaging [Figure 1c] showed a 28-mm giant aneurysm with severe calcification and perifocaledema. Three-dimensional CT angiography (3D-CTA) [Figure 1d] and threedimensional digital subtraction angiography (DSA) of the left internal carotid artery (ICA) [Figure 1e] demonstrated that the aneurysm was located at the left PerA–CMA junction. These examinations showed the presence of five additional aneurysms, including an anterior communicating artery (AcomA) aneurysm and a right middle cerebral artery (MCA) aneurysm.

#### Surgical strategy

Because of the obvious enlargement of the aneurysm and perifocaledema, we planned surgical treatment. Standard neck clipping [Figure 2a] seemed difficult because of severe calcification, similar to that observed in the previous operation. Therefore, three possible strategies were considered for bypass surgery before

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Figure 1: Follow-up images of the aneurysm. (a) Axial computed tomography scan before the first operation in 2003 showing a 15-mm heterogeneous mass in the left frontal lobe. (b) Axial computed tomography scan on admission in 2010 showing a 28-mm giant aneurysm with perifocaledema. The aneurysm is growing up with calcification. (c) Preoperative axial T2-weighted magnetic resonance imaging showing clear high signal intensity of marked perifocaledema in the white matter around the aneurysm. (d) The anteroposterior view of preoperative 3-dimensional computed tomography angiography showing three aneurysms, including a distal anterior cerebral artery aneurysm, an anterior communicating artery aneurysm (short arrow) and a right middle cerebral artery aneurysm (long arrow). (e) The left oblique view of preoperative 3-dimensional digital subtraction angiography of the left internal carotid artery showing the aneurysm at the pericallosal artery (PerA, short arrow)-callosomarginal artery (CMA, long arrow) junction

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operation [Figure 2b–d]. Reconstruction of the left CMA was necessary because it is a major branch supplying the anterior cerebral artery (ACA) territory. However, the possible need for reconstruction of the left distal PerA was left to be decided during operation.

#### Operation

Intraoperative DSA and transcranial motor-evoked potential (MEP) monitoring were performed in this operation. Bifrontal craniotomy was performed on the patient under general anesthesia. Initially, the neck of an AcomA aneurysm was clipped using the interhemispheric approach. We proceeded distally along both A2 vessels and found the DACA aneurysm at the left PerA–CMA junction. The aneurysm was yellowish and solid because of severe calcification [Figure 3a].The left distal PerA was running behind the aneurysm and was densely adherent to the aneurysm because of fibrosis from the previous wrapping operation and could not therefore be dissected from the aneurysm. An occlusion test of the left A2 segment at its proximal portion was performed for 20 min. During this procedure, intraoperative DSA confirmed good retrograde flow in the left distal PerA, and MEP amplitude showed no changes. Based on these findings, the left distal PerA was cut just distal to the aneurysm after clipping without any reconstruction.

Temporary clips were placed on the left A2 segment and left CMA to trap the aneurysm. The aneurysm was incised and the thrombus within the aneurysm was removed. After removal of the thrombus, three lumens were visualized in the aneurysm [Figure 3b]. Neck clipping with reconstruction was attempted but could not be performed without occlusion of the parent artery because of severe calcification of the neck. MEP amplitude subsequently decreased to 50% of the control level while attempting neck clipping. At this point, it was apparent that the neck of the aneurysm could not be clipped with preservation of the feeding artery. Therefore, aneurysm resection and proximal PerA–CMA end-toend anastomosis [Figure 2c] were performed [Figure 3c]. After anastomosis, intraoperative DSA and microvascular



Figure 2: Illustrations showing four possible surgical strategies. Reconstruction for the left distal PerA was to be decided during operation. (b) Shows the reconstructive procedure for the left distal PerA, and (c) and (d) show the surgical strategies requiring no reconstruction. (a) Neck clipping. (b) Left A2 trapping plus A3–A3 side-to-side anastomosis with reconstruction of the left distal PerA by right CMA–left distal PerA side-to-end anastomosis. (c) Left A2 trapping plus bonnet bypass using a short graft of the right superficial temporal artery. (d) Aneurysm resection plus proximal PerA–CMA end-to-end anastomosis



Figure 3: Intraoperative photographs taken during the interhemispheric approach. (a) A yellowish solid aneurysm (box) at the left PerA (short arrow)–CMA (long arrow) junction. (b) Thrombectomy of the aneurysm while trapping the aneurysm after cutting the left distal PerA. The neck of the aneurysm is highly calcified. (c) Left proximal PerA–CMA end-to-end anastomosis



Figure 4: Intraoperative digital subtraction angiography of the left internal carotid artery (oblique views). (a) Left internal carotid angiogram showing the aneurysm at the left PerA (short arrow)–CMA (long arrow) junction. (b) Left internal carotid angiogram under occlusion of the leftA2 segment, showing good retrograde flow to the left distal PerA (arrow) from the left middle cerebral artery through leptomeningeal anastomosis (arrow). (c) Left internal carotid angiogram after aneurysm resection and proximal PerA–CMA end-to-end anastomosis, showing elimination of the distal anterior cerebral artery aneurysm (arrow head) and good recanalization of the left CMA (arrow)

Doppler ultrasonography demonstrated good flow in the left CMA and adequate retrograde flow to the left distal PerA from the left MCA through leptomeningeal anastomosis. MEP amplitude was maintained at the control level until dural closure. Findings from intraoperative DSA are shown in Figure 4.

#### **Postoperative course**

The patient had no neurological deficits after operation, and the postoperative CT scan demonstrated no infarction including the left ACA territory [Figure 5a]. Elimination of the aneurysm and good flow in the left CMA were observed on 3D-CTA [Figure 5b].The patient was doing well 6 months after operation.

#### **DISCUSSION**

Giant intracranial aneurysms (>25 mm in diameter) comprise approximately 5% of all intracranial aneurysms,<sup>[1,2,5,22]</sup> and most of them arise on the proximal



Figure 5: Postoperative computed tomography scan and 3-dimensional CT angiography. (a) Computed tomography scan after operation showing no ischemic lesion in the left anterior cerebral artery territory. (b) The oblique view of postoperative 3-dimensional CT angiography showing elimination of the aneurysm and good recanalization of the left callosomarginal artery (arrow)

segment of the cerebral arteries, such as ICAs and the vertebrobasilar arteries. In most cases, standard neck clipping is difficult because of specific characteristics of the aneurysms, such as the broad neck, involvement of the branch vessels, calcification and thrombus. In the cases of giant ACA aneurysms, Yokoh *et al.* studied and proposed many variations of bypass surgeries to reconstruct ACAs and their branches.<sup>[36]</sup> The aneurysms should be treated using appropriate surgical methods because of their poor natural history.

Although the natural history of unruptured giant intracranial aneurysms has not been sufficiently clarified, many authors reported high rupture rates and high mortality.<sup>[1,2,16,22,33]</sup> The risk of aneurysm rupture increases with aneurysm size. In the ISUIA (International Study of Unruptured Intracranial Aneurysms), the 5-year cumulative rupture rate for anterior circulation was 40% and that for posterior circulation was 50%.<sup>[33]</sup> Peerless *et al.*, who reported 31 untreated cases of patients with giant aneurysms, including 25 saccular and 6 fusiform types, stated that the mortality rate with the saccular type was 68% at 2 years and that all but four patients were dead at 5 years.<sup>[22]</sup> Moreover, when an aneurysm is extensively or completely thrombosed, it still has a risk of rupture and the natural history may be even poorer.<sup>[2]</sup>

Most DACA aneurysms arise at the PerA–CMA junction.<sup>[14]</sup> In de Sousa's report, 61 of 74 aneurysms were located at the PerA–CMA junction.<sup>[4]</sup> Yasargil stated that DACA aneurysms present specific difficulties during surgery, including a narrow working space in the interhemispheric fissure, dense adhesions between the cingulated gyri, a broad-based and/or sclerotic neck in the aneurysm, difficulty in controlling the parent artery, the fixed dome on the pial layer and increased association of multiple aneurysms and vascular anomalies.<sup>[35]</sup> In addition, they are fragile and frequently rupture prematurely during exposure.

#### Table 1: Summary of reported cases of giant distal anterior cerebral artery aneurysms

Author (year)	Age	Sex	Symptoms and signs	Treatment	Outcome
Drake CG (1979)	41	Μ	SAH	Occlusion of the neck, evacuation of the thrombus	Dead
Pia HW <i>et al</i> . (1979)	-	-	SAH	Clipping of the feeding artery, resection of the sac	Good
O'neill M <i>et al.</i> (1980)	29	F	Loss of limb control, headache, ataxia, incontinence	Thrombectomy	Good
Pozzati E <i>et al</i> . (1982)	20	F	Headache, papilledema	Clipping of the feeding artery, aneurysm resection	Good
Smith RR <i>et al</i> . (1982)	67	Μ	SAH	Aneurysm resection, end-to-end anastomosis of ACA	Good
Shigemori M <i>et al.</i> (1982)	69	F	Headache, hemiparesis, memory disturbance, disorientation	Neck clipping, thrombectomy	Good
Hayashi M <i>et al</i> . (1985)	57	Μ	Transient hemiparesis	Conservative	Unchanged
Hayashi M <i>et al</i> . (1985)	59	Μ	Seizures, anosmia	Neck clipping, thrombectomy	Good
Yamagami T <i>et al</i> . (1986)	51	Μ	Gait disturbance, dysarthria, dysphasia, left to right aqnosia	Neck clipping	Good
Nitta T <i>et al</i> . (1987)	61	М	Headache	Thrombectomy, aneurysmorrhaphy	Good
Fukushima T (1987)	68	М	No rupture	Neck clipping	Dead
Maiuri F <i>et al</i> . (1990)	64	M	Transient ataxia, loss of	Conservative	Unchanged
			vision, dizziness		
Mishima K <i>et al</i> . (1990)	53	Μ	Meningitis	Neck clipping, thrombectomy	Vegetative
Hernesniemi J et al. (1992)	-	-	-	_	Good
Preul M <i>et al</i> . (1992)	72	М	Mental deterioration	<ul> <li>Clipping of the feeding artery, thrombectomy</li> </ul>	Dysphasia
Hashizume K et al. (1992)	67	М	SDH	Neck clipping, resection of the sac	Good
Shiokawa K <i>et al</i> . (1993)	69	F	SAH	Neck clipping, thrombectomy	Good
Farias JP et al. (1997)	49	F	Intracerebral hematoma	Neck clipping, thrombectomy	Good
de Sousa AA et al. (1999)	-	-	-	Operation (clipping)	Good
de Sousa AA <i>et al.</i> (1999)	-	-	-	Operation (clipping)	Good
Kanemoto Y <i>et al</i> . (2000)	77	F	Acute onset of akinetic	Neck clipping, thrombectomy	Good
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Ewald CH <i>et al</i> . (2000)	-	-	Headache, hemiparesis	Clipping of the feeding artery, CCA–PerA side-to- end anastomosis using a saphenous vein graft	Good
Koyama S (2000)	66	F	SAH, SDH	Neck clipping	Dead
Hoh BL et al. (2001)	-	-	-	Endvascular proximal occlusion	Good
Hoh BL <i>et al</i> . (2001)	-	-	-	Endovascular proximal occlusion, A3–A3 side-to- side anastomosis	Good
Ture U <i>et al</i> . (2001)	65	F	Headache, personality changes, seizures, papilledema	Neck clipping, thrombectomy	Good
Topsakal C <i>et al</i> . (2003)	65	F	SAH	Neck clipping	Dead
Biondi A et al. (2006)	60	F	Visual deficit	Endovascular proximal occlusion	Good
Biondi A <i>et al</i> . (2006)	49	Μ	SAH	Endovascular proximal occlusion	Good
Steven DA et al. (2007)	-	-	-	Operation	-
Steven DA et al. (2007)	-	-	-	Operation	-
Park DH et al. (2008)	65	F	Headache, memory loss, hemiparesis	Neck clipping, thrombectomy	Good
Matsushima K <i>et al</i> . (2011)	69	F	None	Aneurysm resection, proximal PerA–CMA end-to- end anastomosis	Good

Gray zone of the table indicates four cases treated with bypass surgery, SAH: Subarachnoid hemorrhage, ACA: Anterior cerebral artery, SDH: Subdural hematoma, CCA: Common carotid artery, PerA: Pericallosal artery, CMA: Callosomarginal artery

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Giant DACA aneurysms are extremely rare. This may be due to their tendency to rupture early. This feature may be related to anatomical features such as the lack of resistant arachnoid membranes at the level of the pericallosal cisterns.<sup>[28]</sup> In 2009, Gelfenbeyn et al. reviewed the largest series of 26 cases of giant DACA aneurysms.<sup>[8]</sup> We reported here six more cases<sup>[3,12,13,21]</sup> and one additional case of our own, thereby reviewing a total of 33  $cases^{[3-13,15,17-21,23-27,29-32,34]}$  [Table 1]. Thirty of the 33 cases were treated surgically, and the neck was clipped in 13 of them.<sup>[7,9,10,13,15,18,21,26,27,31,32,34]</sup> Only four cases were treated with bypass surgery, and good outcomes were obtained in all of them. The first bypass surgery, including aneurysm resection and end-to-end anastomosis of ACA, was reported in 1982.<sup>[29]</sup> The second case involved clipping of the parent artery and common carotid artery-PerA sideto-end anastomosis using a saphenous vein graft,<sup>[6]</sup> while the third case involved A3-A3 side-to-side anastomosis with endovascular proximal occlusion.<sup>[12]</sup>

In our case, standard neck clipping with preservation of the parent artery was impossible because of severe calcification. A3–A3 side-to-side anastomosis seemed difficult in the narrow and deep working space in the interhemispheric fissure, and it might have required longer occlusion time. Furthermore, the bonnet bypass using the right superficial temporal artery was suspected to cause ischemia of the scalp in our case. Eventually, PerA–CMA end-to-end anastomosis with aneurysm resection was selected as the most appropriate treatment.

End-to-end anastomosis with aneurysm resection is usually excluded as a treatment for aneurysms because the vessels cannot be anastomosed as a result of their lengths. However, in some cases of aneurysm at the PerA– CMA junction, adequate retrograde flow in the distal PerA can be expected because PerA is supplied from the posterior cerebral artery through the posterior PerA or from MCA through leptomeningeal anastomosis. In such cases, reconstruction of the distal PerA is not necessary. Moreover, when the distal PerA is cut, proximal PerA– CMA end-to-end anastomosis becomes easier because the anastomosis site becomes more flexible and tension at this site reduces.

For safe treatment of giant aneurysms, it is very important to plan alternative strategies before operation and change the strategy during operation if necessary. Moreover, intraoperative modalities including intraoperative DSA and MEP monitoring are helpful in deciding the strategy. In our case, the distal PerA could be cut without any reconstruction because adequate retrograde flow was observed in DSA. The decreased MEP amplitude suggested occlusion of the parent artery, and the recovery of MEP amplitude suggested preservation of the parent artery. After anastomosis, intraoperative DSA and microvascular Doppler ultrasonography proved good patency of anastomosis.

#### CONCLUSION

Giant DACA aneurysms are extremely rare, and surgical treatment is very difficult in most cases. We conclude that proximal PerA–CMA end-to-end anastomosis with aneurysm resection is a useful treatment for thrombosed giant DACA aneurysms at the PerA–CMA junction, especially when the distal PerA can be cut. In this report, we also presented the effectiveness of intraoperative modalities, such as intraoperative DSA and MEP monitoring, for performing a safe operation.

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