

Diagnosis and Operative Management of Carotid Endarterectomy in Patients with Twisted Carotid Bifurcation

Masaaki UNO,¹ Kenji YAGI,¹ Hiroyuki TAKAI,¹ Keijiro HARA,¹
Naoki OYAMA,² Yoshiki YAGITA,² and Shunji MATSUBARA¹

¹Department of Neurosurgery, Kawasaki Medical School, Kurashiki, Okayama, Japan

²Department of Stroke Medicine, Kawasaki Medical School, Kurashiki, Okayama, Japan

Abstract

Although carotid endarterectomy (CEA) is an established procedure, technical modifications are required when anatomical features are unusual. The present study aimed to determine the characteristics of diagnostic features, surgical management, and outcomes of patients with a twisted carotid bifurcation (TCB). We assessed 108 consecutive patients by cervical carotid echography (CCE) and black-blood magnetic resonance imaging (BB-MRI) before they underwent 115 CEA procedures. We classified carotid bifurcation (CB) anatomy based on anteroposterior findings of the internal carotid artery (ICA) and external carotid artery (ECA) determined by cerebral or three-dimensional computed tomographic angiography as follows. The ICA and ECA ran laterally and medially, respectively, in Type 1, overlapped in Type 2, and the ICA and ECA ran medially and laterally, respectively, in Type 3. We also classified the patients according to whether or not they had a TCB and compared their diagnostic findings, clinical characteristics, and surgical outcomes. The numbers of patients with Types 1, 2, and 3 were 74 (64.4%), 32 (27.8%), and 9 (7.8%), respectively, and 13 (11.3%) with a TCB included four patients with Type 2 and all nine patients with Type 3. The appearance of Type 3 differed from that of the other two types on CCE and BB-MR images. After correcting the anatomical location of a TCB, surgical duration and adverse event rates did not significantly differ between patients with and without a TCB. Patients with a TCB could safely undergo CEA after correcting the ICA to the normal position.

Keywords: carotid endarterectomy, twisted carotid bifurcation, MRI, cervical carotid echography, surgical complications

Introduction

Carotid endarterectomy (CEA) reduces overall stroke risk for symptomatic and asymptomatic patients with severe carotid stenosis.^{1,2} Although CEA is an established procedure, technical modifications are required in the face of unusual anatomical features. The external carotid artery (ECA) is usually located medially to the ipsilateral internal carotid artery (ICA) at the level of the common carotid artery (CCA) bifurcation. However, the ICA sometimes runs medially to the ECA, which has been described as side-by-side carotid artery,³ dorsal/dorsomedial origin of the ICA,⁴ complete transposition of the carotid bifurcation (CB),⁵ twisted

carotid bifurcation (TCB),^{6–8} lateral ECA, and lateral position of the ECA (LPECA).^{9–12} Most recent reports of patients with TCB who have undergone CEA have focused on surgical techniques, and few describe diagnosing a TCB.^{8,10} The present study aimed to determine the diagnostic features of a TCB and its surgical management and outcomes.

Materials and Methods

A total of 108 patients (mean age, 69.9 ± 6.9 years; 12 females) underwent 115 consecutive CEA procedures at our institution between April 2009 and November 2018. Patients were assessed by cervical carotid echography (CCE) and black-blood magnetic resonance imaging (BB-MRI) before undergoing CEA. All patients were assessed by conventional angiography (digital subtraction angiography [DSA]) or three-dimensional computed tomographic angiography (3D-CTA) to diagnose ICA stenosis.

Received February 15, 2020; Accepted April 21, 2020

Copyright© 2020 by The Japan Neurosurgical Society This work is licensed under a Creative Commons Attribution-NonCommercial-NoDerivatives International License.

The surgical indications for CEA for asymptomatic and symptomatic patients were defined as >60% and >50% stenosis, respectively,^{1,2)} and for symptomatic patients with vulnerable plaque as <50% stenosis after discussion at a stroke conference.¹³⁾

Figure 1 shows the anatomy of the CB on anteroposterior views of DSA or 3D-CTA classified according to Kamide as Types 1 to 3, in which the ICA and ECA run laterally and medially, respectively (Type 1; Fig. 1A), overlap (Type 2; Fig. 1B), and run medially and laterally, respectively (Type 3; Fig. 1C).⁶⁾ We also categorized the patients according to whether or not they had a TCB and compared the diagnostic findings, clinical characteristics, and surgical outcomes. A TCB was defined as the ECA lying posterior or posterolateral to the ICA in the operative field. We corrected the position of the ICA to the lateral side of the ECA before proceeding with CEA.⁶⁻⁸⁾

The baseline conditions of patients were defined as follows. Hypertension was defined as systolic and/or diastolic blood pressure of ≥ 140 and ≥ 90 mmHg, respectively, before CEA or use of antihypertensive agents. Diabetes mellitus was defined as glycosylated hemoglobin A1c >6.5% or use of hypoglycemic agents. Hyperlipidemia was defined as low-density lipoprotein (LDL)-cholesterol >140 mg/dL or use of antihyperlipidemic agents. Ischemic heart disease was defined as a history of angina pectoris, myocardial infarction, or use of antiplatelet agents for coronary artery disease.

Rates of carotid stenosis were calculated according to the North American Symptomatic Carotid Endarterectomy Trial (NASCET).²⁾

CCE proceeded using HI VISION Preirus (Hitachi Ltd., Tokyo, Japan) or Xario XG (Toshiba Medical Systems Corp., Tochigi, Japan) equipment, with a linear probe used by two experts (NO and YY; Figs. 1D–1F).

The BB-MRI system included a 1.5 T MRI machine with a Signa Excite High Speed Scanner (General Electric, Milwaukee, WI, USA). Two authors (HT and NO) assessed the anatomical locations of the ICA and ECA, and the signal intensity and location of plaque on T1-weighted axial MR images. We classified the views of CB on BB-MR axial images according to whether the ICA was located posterolaterally from the midline of the ECA (Type 1; Fig. 1G), did not deviate from side to side outside the midline of the ECA (Type 2; Fig. 1H), or was located posteromedially from the midline of the ECA (Type 3; Fig. 1I). We also classified the locations of plaque on T1-weighted axial images into four regions (Figs. 2A and 2B).

The adverse events of stroke, cranial nerve palsy, wound hematoma, and cerebral hyperperfusion syndrome were also evaluated at 90 days after

procedures. The first author (MU) implemented or supervised all procedures.

The Ethics Committee at our institution approved the protocol of this retrospective study and waived the requirement for patient consent (No. 3529).

Statistical analysis

Numerical data and categorical variables are expressed as means \pm standard deviation (SD) and as numbers (n) with ratios (%), respectively. Data were statistically analyzed using SPSS version 24 software (IBM Corp., Tokyo, Japan). χ^2 tests, Fisher's extraction tests, analyses of variance (ANOVA), and a regression model were used. Variables with $p < 0.20$ in univariable logistic regression analyses were included in multivariable logistic regression analyses to assess independent predictors. Values with $p < 0.05$ were considered statistically significant.

Results

Table 1 shows the relationships between CEA and age, sex, vascular risk factors, rates of TCB, and outcomes of each type. Age >70 years, sex, number of risk factors, being symptomatic, and degree of ICA stenosis did not differ among the groups. However, in 18 of 32 CEA for Type 2 and in all 9 for Type 3, CB were located on the right side. Of 13 (11.3%) patients with a TCB who were treated by CEA, 4 (12.5%) had Type 2 and 9 (100%) had Type 3. The TCB were located on the right and left in 11 (84.6%; $p = 0.02$) and 2 (15.4%) of these patients, respectively (Tables 1 and 2). Among female patients, 3 (23.1%, $p = 0.14$) had a TCB. The clinical risk factors of hypertension, diabetes mellitus, hyperlipidemia, and ischemic heart disease did not significantly differ among the three types of CB or between patients with and without a TCB (Tables 1 and 2).

Findings of CCE and BB-MRI

All patients except one were assessed by CCE before CEA. The findings showed that the anatomical features considerably differed between Type 3 and Types 1 and 2. The ECA was visualized on the surface, and ICA was located below the ECA in a Y shape, and the ICA was identified by the flow pattern (Fig. 1F).

Most ($n = 101$) patients were also assessed by BB-MRI, which defined positional relationships between the ICA and the ECA (Figs. 1G–1I). An ICA that contained high-intensity plaque was obvious, running vertically, and deeply to the ECA in Type 2 (Fig. 1H) and deeply and medially to the ECA in all Type 3 (Fig. 1I).

Plaque was mainly located in the posteromedial side (region 2) in 90% of patients with a TCB. In

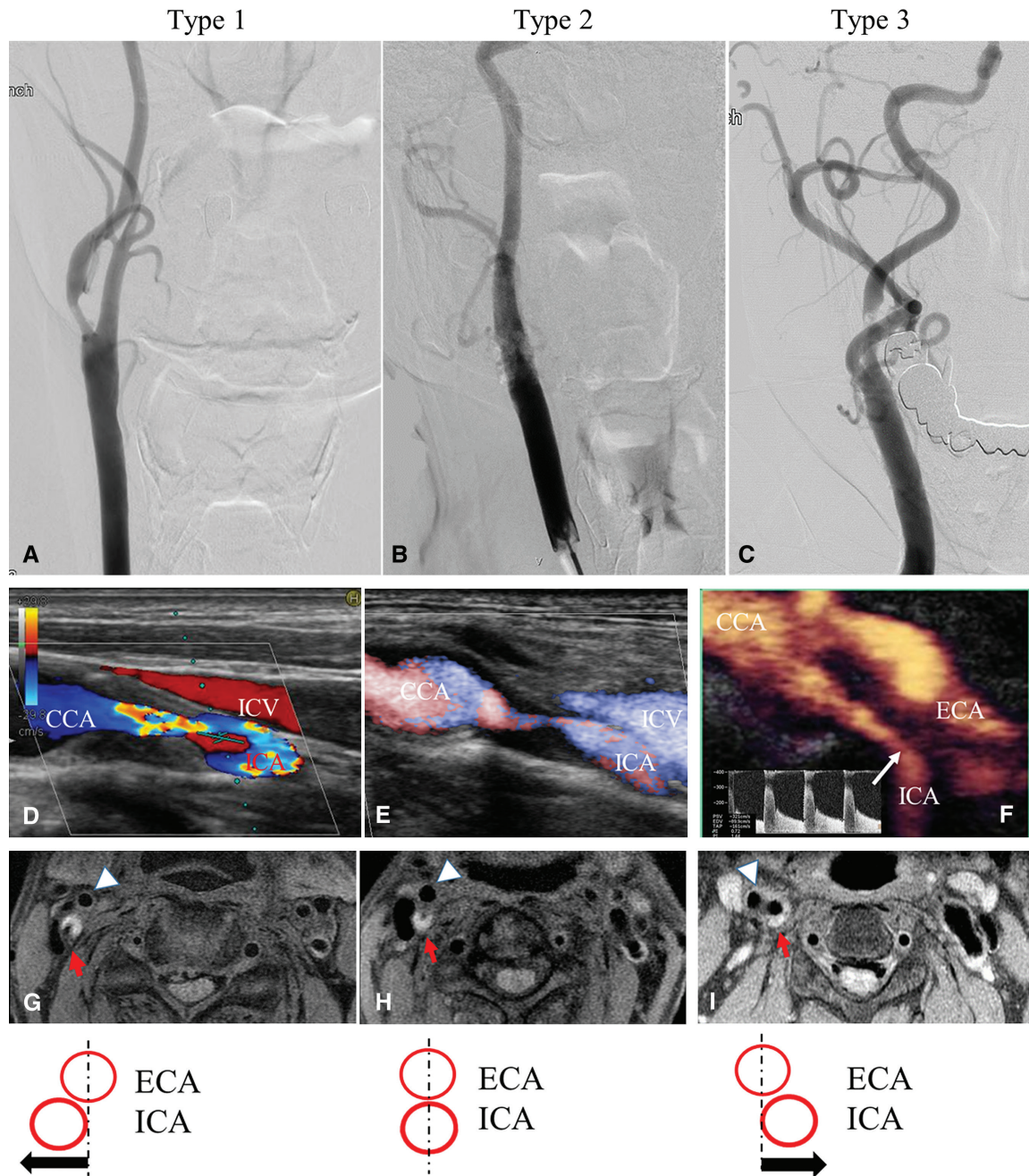


Fig. 1 Anatomy of carotid bifurcations classified according to Kamide as Types 1–3. Anteroposterior views of ICA and ECA on digital subtraction angiograms. All carotid bifurcations were classified as follows. (A) Type 1: ICA and ECA run laterally and medially, respectively. (B) Type 2: ICA and ECA overlap. (C) Type 3: ICA and ECA run medially and laterally, respectively. Preoperative CCE and BB-MRI findings of each type; CCE images of Types 1 (D), 2 (E), and 3 (F). The appearance of Type 3 differed from those of Types 1 or 2 in that the ECA is visible on the surface, and the ICA is located below the ECA in a Y shape. High flow with low resistance in the ICA was confirmed by broad systolic peaks and high end-diastolic flow velocity (F). BB-MRI of Types 1 (G), 2 (H), and 3 (I). High-intensity plaque in the ICA located laterally to the ECA in Type 1 (G), vertically and deeply to the ECA in Type 2 (H) and deeply medial to the ECA in Type 3 (I). Red arrow, ICA; white triangle, ECA. BB-MRI: black-blood magnetic resonance imaging, CCA: common carotid artery, CCE: cervical carotid echography, ECA: external carotid artery, ICA: internal carotid artery.

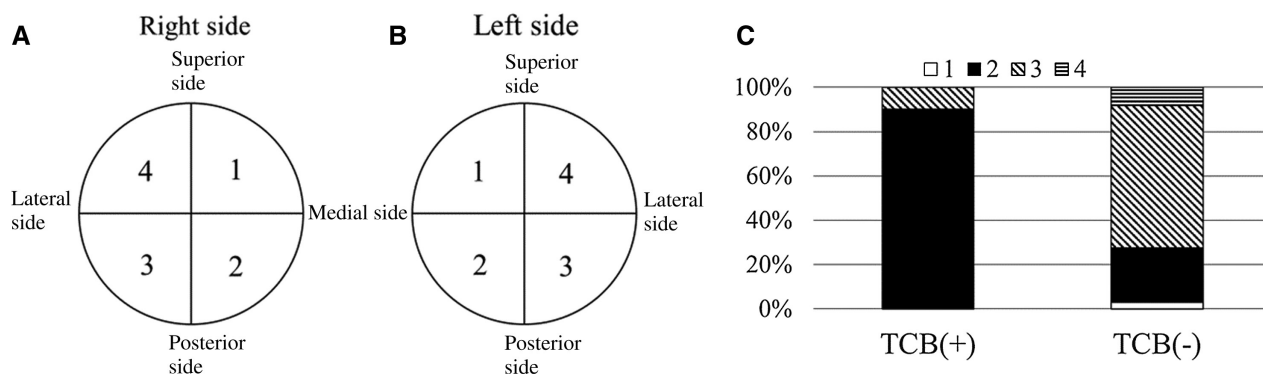


Fig. 2 Locations of plaque on T1-weighted axial MR images. Right (A) and left (B) locations. Left (C) and right (D) bars, locations in patients with and without TCB, respectively. MR: magnetic resonance, TCB: twisted carotid bifurcation.

Table 1 Relationships between CEA and age, sex, vascular risk factors, rates of TCB, and the outcomes of each type

	Type 1 N = 74 (66.4%)	Type 2 N = 32 (27.8%)	Type 3 N = 9 (7.8%)	p value
Age ≥ 70 y.o.	49 (66.2%)	17 (53.1%)	5 (55.6%)	0.41
Female	8 (10.8%)	2 (6.3%)	2 (22.2%)	0.38
Hypertension	61 (82.4%)	25 (78.1%)	7 (77.8%)	0.85
DM	40 (54.1%)	14 (43.8%)	2 (22.2%)	0.16
Hyperlipidemia	44 (59.5%)	16 (50.0%)	4 (44.4%)	0.52
IHD	14 (18.9%)	4 (12.5%)	0 (0%)	0.14
Symptomatic	57 (77.0%)	21 (65.6%)	9 (100%)	0.09
ICA stenosis $\geq 70\%$	41 (55.4%)	19 (59.4%)	5 (55.6%)	0.93
Right side lesion	34 (45.9%)	18 (56.3%)	9 (100%)	0.008
Twisted CB in CEA	0 (0%)	4 (12.5%)	9 (100%)	<0.001
Operation time (min) ≥ 4 hours	26 (35.1%)	12 (37.5%)	4 (44.4%)	0.85
Adverse event	7 (9.5%)	1 (3.1%)	1 (11.1%)	0.5

CB: carotid bifurcation, CEA: carotid endarterectomy, DM: diabetes mellitus, ICA: internal carotid artery, IHD: ischemic heart disease.

contrast, plaque was mainly located in the postero-lateral side (region 3) in 63 (64.3%) of 98 CEA procedures in patients without a TCB (Fig. 2C). The ratio of high plaque intensity (>1.5 compared with the sternocleidomastoid muscle) measured by T1-weighted axial MRI between patients with and without a TCB did not significantly differ (30.0% vs. 36.7%, respectively, $p = 0.517$).

Surgical procedure for patients with TCB

The neck of the patient was extended under general anesthesia and rotated approximately 45° to the contralateral side of the lesion. A Z-shaped skin incision was selected for cosmetic reasons. When a TCB was predicted by preoperative

examination, the head was rotated approximately 60° to the contralateral side of the lesion. The ECA was dissected farther for patients with than without a TCB (Fig. 3A). The ICA lying behind the ECA was dissected and its carotid sheath was raised to the lateral side using a hook with a rubber loop (Fig. 3B). The ICA was then moved to the normal anatomical position (Fig. 3C). All TCB in 13 patients were surgically manipulated in this manner.

The ICA was located at the CB behind the ECA in four patients with Type 2 CB that were judged as twisted. However, the ECA ran across the ICA at the distal site, so the ECA required longer dissection to lift the ICA. We therefore confirmed TCB in these four patients.

Table 2 Relationships between CEA and age, sex, vascular risk factors, and the outcomes of patients with non-twisted CB and with twisted CB

	Non-twisted CB N = 102	Twisted CB N = 13	p value
Age \geq 70 y.o.	63 (61.8%)	8 (61.5%)	>0.99
Female	9 (8.8%)	3 (23.1%)	0.14
Hypertension	84 (82.4%)	9 (69.2%)	0.27
DM	53 (52.0%)	3 (21.1%)	0.08
Hyperlipidemia	59 (57.8%)	5 (38.5%)	0.24
IHD	18 (17.6%)	0 (0%)	0.22
Symptomatic	76 (74.5%)	11(84.6%)	0.73
ICA stenosis \geq 70%	58 (56.9%)	7 (53.8%)	>0.99
Right side	50 (49.0%)	11 (84.6%)	0.02
Operation time (min) \geq 4 hours	36 (35.3%)	6 (46.2%)	0.54
Adverse event	7 (6.9%)	2 (15.4%)	0.27

CB: carotid bifurcation, CEA: carotid endarterectomy, DM: diabetes mellitus, ICA: internal carotid artery, IHD: ischemic heart disease.

Surgical duration

The surgical durations for Types 1, 2, and 3 were 222.6 ± 35.4 , 231.0 ± 37.4 , and 244.3 ± 36.6 min, respectively. Procedural length did not significantly differ according to type ($p = 0.17$; ANOVA). Similarly, the surgical duration and ratio of >4 hours of surgery between the groups with and without a TCB did not significantly differ (Table 2).

Surgical complications

One patient with a TCB developed a temporal neurological deficit and another developed a wound hematoma. However, postoperative diffusion-weighted MRI (DWI) did not reveal any new abnormalities in patients with a TCB. In contrast, cerebral hyperperfusion syndrome ($n = 1$), cerebral ischemia ($n = 3$), cranial nerve palsy ($n = 2$), and wound hematoma ($n = 1$) developed and postoperative DWI revealed new abnormalities in seven (6.9%) patients without a TCB. Complication rates including temporal deficits between patients with and without a TCB and among the three types of CB did not significantly differ (15.4% vs. 6.9%, $p = 0.27$; Table 2 and $p = 0.5$; Table 1, respectively).

Discussion

Criteria for TCB have not been defined. Therefore, we defined criteria for TCB according to whether the ECA lay posteriorly or posterolaterally to the ICA in the operative field and whether moving the ICA to the correct lateral side of the ECA was required.^{7,8)} The frequency of TCB among our patients

was 11.3% and it was significantly more prevalent on the right side. Angiography findings have shown that the incidence of TCB or LPECA in various diseases is 4.3%–4.9% in Japanese populations^{10,12)} and 3.6%–15.1% during CEA.^{5–8,10,14,15)} Therefore, this abnormality is actually quite prevalent.

Although a scientific explanation for the right-sided dominance of TCB has not yet been supported by convincing evidence, congenital and arteriosclerosis hypotheses have been proposed.^{6–9,11)} The congenital hypothesis proposes that TCB is a result of excessive mediolateral migration of the ECA during embryogenesis.^{9,11)} The other hypothesis proposes that elongation and tortuosity of the carotid artery are due to age-related arteriosclerosis.^{6,7)}

Diagnosis of TCB

The anatomical status of the ECA and ICA is important to determine before patients undergo CEA using CCE and BB-MRI and to confirm it using angiography in the anteroposterior view. The original purpose of CCE and BB-MRI was to determine the vulnerability of carotid plaque and degree of stenosis. However, structural abnormalities of the ECA and ICA should be identified by CCE. The ECA of patients with Type 3 CB was visualized on the surface, and the ICA appeared below the ECA in a Y shape. Wave patterns can differentiate the ECA from the ICA.

Sitzer et al.⁴⁾ defined a dorsal/dorsomedial origin of the ICA as an angle of $\geq 60^\circ$. Some patients with Type 2 and all with Type 3 CB had this type of origin. They also found that a dorsal/dorsomedial

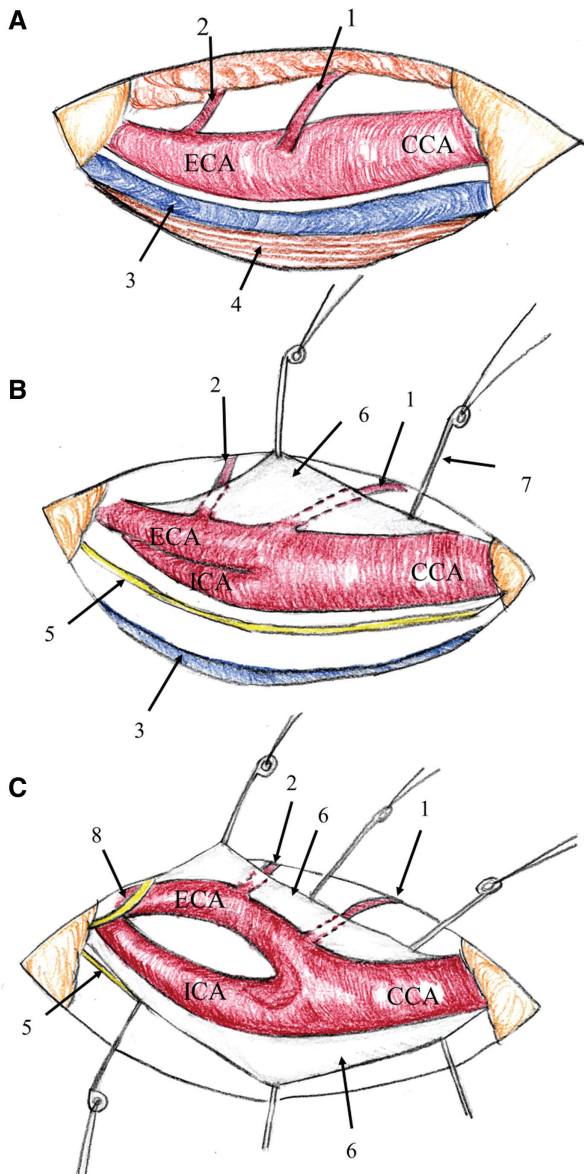


Fig. 3 Schematic illustration of the steps required to dissect the right ICA in patients with TCB. (A) Initially, the ICA was not identified. Therefore, the ECA was further dissected to the distal side. (B) After lifting the ECA medially, the ICA at the bifurcation was identified, dissected, and moved laterally using hooks with rubber loops. (C) The dissected ICA was lifted laterally and superficially using hooks with rubber loops. Hooks were also used to pull out the sternocleidomastoid muscle that hangs over the ICA and lift the carotid sheath and connective tissues around the CCA, ICA, and ECA. 1, Superior thyroid artery; 2, lingual artery; 3, internal jugular vein; 4, sternocleidomastoid muscle; 5, vagus nerve; 6, carotid sheath; 7, hook with a rubber loop, 8, hypoglossal nerve. CCA: common carotid artery, ECA: external carotid artery, ICA: internal carotid artery, TCB: twisted carotid bifurcation.

origin of the ICA was significantly more prevalent on the right side.⁴⁾ However, the anatomical relationships between the ECA and ICA of patients scheduled to undergo CEA have never been evaluated by CCE.⁵⁾

The ICA location can be easily confirmed by BB-MRI because its diameter is larger than that of the ECA. Furthermore, plaque in the ICA frequently presents as hyperintense regions on BB-T1-weighted MRI. Katano et al. have determined TCB using CTA,⁶⁾ but BB-MRI assessment of TCB in patients scheduled to undergo CEA has not been reported.

Although plaque intensity on BB-MRI did not differ between patients with and without a TCB, the posteromedial location of plaque was more prevalent in patients with than without a TCB.

We emphasize that non-invasive evaluations such as CCE and BB-MRI can precisely identify relationships between the ECA and ICA in addition to the features of carotid plaque.

Surgery for TCB

All patients with a TCB safely underwent CEA after ICA transposition to the normal anatomical location. The key issues of CEA under these circumstances were as follows. The head and neck were turned radically to the contralateral side, the ECA was dissected beyond the CB to find the ICA behind it, the ICA was dissected, and the carotid sheath was lifted using a hook with a rubber loop to shift the ICA more laterally and superficially. Toyota et al.¹⁶⁾ describe the use of a Lone Star retractor system during CEA. We have applied the same principle using hooks with a rubber loop to lift the ECA and the ICA during routine CEA. This type of hook is very useful in CEA for TCB as it can also pull out the sternocleidomastoid muscle that hangs over the ICA.

Previous reports have described similar procedures for patients who are scheduled to undergo subsequent TCB.^{7,8,14)} However, Kamide et al. used another approach for such patients. They operated up the medial, rather than the lateral side of the carotid tree.⁶⁾ Loftus commented on the difficulty of suturing and exposing the distal site of the ICA during this approach and recommended transposing the ICA to the normal location^{14,17)} The surgical duration tended to increase according to the presence/absence and type of TCB, but the difference did not reach significance. The ICA must be gently manipulated to protect against distal embolisms arising from plaque. Adverse event rates and new postoperative lesions on DWI did not significantly differ between patients with and without a TCB when the surgical modification described herein was applied. Surgeons should understand carotid abnormalities and create

an appropriate preoperative strategy to ensure the safety of CEA for patients with a TCB.

Conclusions

The incidence of TCB among 108 patients who underwent CEA at our institution was 11.3%. A TCB can be identified by CCE and BB-MRI before undergoing CEA. Patients with a TCB could safely undergo CEA after transposing the ICA to the normal location.

Conflicts of Interest Disclosure

The authors have no conflicts of interest to declare regarding this study or its findings.

References

- 1) Endarterectomy for asymptomatic carotid artery stenosis: Executive committee for the asymptomatic carotid atherosclerosis study. *JAMA* 273: 1421–1428, 1995
- 2) North American Symptomatic Carotid Endarterectomy Trial Collaborators, Barnett HJM, Taylor DW, Haynes RB, et al.: Beneficial effect of carotid endarterectomy in symptomatic patients with high-grade carotid stenosis. *N Engl J Med* 325: 445–453, 1991
- 3) Loftus CM: Side by Side Carotid Endarterectomy. St. Louis: Principles and Technique Quality Medical Publishing, pp 48–49, 1995
- 4) Sitzler M, Puac D, Buehler A, et al.: Internal carotid artery angle of origin: a novel risk factor for early carotid atherosclerosis. *Stroke* 34: 950–955, 2003
- 5) Marcucci G, Accrocca F, Gabrielli R, et al.: Complete transposition of carotid bifurcation: can it be an additional risk factor of injury to the cranial nerves during carotid endarterectomy? *Interact Cardiovasc Thorac Surg* 13: 471–474, 2011
- 6) Kamide T, Nomura M, Tamase A, et al.: Simple classification of carotid bifurcation: is it possible to predict twisted carotid artery during carotid endarterectomy? *Acta Neurochir (Wien)* 158: 2393–2397, 2016
- 7) Katano H, Yamada K: Carotid endarterectomy for stenoses of twisted carotid bifurcations. *World Neurosurg* 73: 147–154; discussion e21, 2010
- 8) Tokugawa J, Kudo K, Mitsuhashi T, Yanagisawa N, Nojiri S, Hishii M: Surgical results of carotid endarterectomy for twisted carotid bifurcation. *World Neurosurg* 2019:S1878–8750(1819)30401–30402, 2019
- 9) Handa J, Matsuda M, Handa H: Lateral position of the external carotid artery. Report of a case. *Radiology* 102: 361–362, 1972
- 10) Ito M, Niiya Y, Kojima M, et al.: Lateral position of the external carotid artery: a rare variation to be recognized during carotid endarterectomy. *Acta Neurochir Suppl* 123: 115–122, 2016
- 11) Teal JS, Rumbaugh CL, Bergeron RT, Segall HD: Lateral position of the external carotid artery: a rare anomaly? *Radiology* 108: 77–81, 1973
- 12) Ueda S, Kohyama Y, Takase K: Peripheral hypoglossal nerve palsy caused by lateral position of the external carotid artery and an abnormally high position of bifurcation of the external and internal carotid arteries—a case report. *Stroke* 15: 736–739, 1984
- 13) Takai H, Uemura J, Yagita Y, et al.: Plaque characteristics of patients with symptomatic mild carotid artery stenosis. *J Stroke Cerebrovasc Dis* 27: 1930–1936, 2018
- 14) Hayashi N, Hori E, Ohtani Y, Ohtani O, Kuwayama N, Endo S: Surgical anatomy of the cervical carotid artery for carotid endarterectomy. *Neurol Med Chir (Tokyo)* 45: 25–29, discussion 30, 2005
- 15) Schulz UG, Rothwell PM: Major variation in carotid bifurcation anatomy: a possible risk factor for plaque development? *Stroke* 32: 2522–2529, 2001
- 16) Toyota S, Kumagai T, Goto T, Mori K, Taki T: Utility of the lone star retractor system in microsurgical carotid endarterectomy. *World Neurosurg* 101: 509–513, 2017
- 17) Loftus CM, Quest DO: Technical issues in carotid artery surgery 1995. *Neurosurgery* 36: 629–647, 1995

Address reprint requests to: Masaaki Uno, MD, Department of Neurosurgery, Kawasaki Medical School, 577 Matsushima, Kurashiki, Okayama 701-0192, Japan
e-mail: muno@med.kawasaki-m.ac.jp