Online December 29, 2020
Journal of Neuroendovascular Therapy 2021; 15: 456–459 DOI: 10.5797/jnet.cr.2020-0188

Case Report

Carotid Artery Stenting for Carotid Arterial Fibromuscular Dysplasia Evaluated Vascular Wall Structure Using Optical Coherence Tomography: A Case Report

Shogo Oshikata,[1] Kei Harada,[2] Masahito Kajihara,[2] Kunihiro Ueta,[1] and Hideki Komatani[1]

Objective: Fibromuscular dysplasia (FMD) is often diagnosed based on angiography. However, it is difficult to distinguish from vasculitis by angiography. Therefore, it is important to evaluate the detailed intravascular findings of lesions in FMD using optical coherence tomography (OCT).

Case Presentation: We present a case of a 30-year-old woman with left carotid artery stenosis. The lesion was diagnosed with a suspected case of FMD by MRA, and gradually progressed over the course of 7 years. Therefore, we underwent carotid artery stenting (CAS) using OCT, and good dilatation of the lesion was obtained.

Conclusion: OCT evaluation during CAS allowed for a definitive diagnosis of FMD because the OCT images revealed a detailed finding of the three layers of the carotid artery.

Keywords ▶ fibromuscular dysplasia, optical coherence tomography, carotid artery stenting

Introduction

Fibromuscular dysplasia (FMD) is an idiopathic, multifocal, non-inflammatory, and non-atherosclerotic disease that affects small- to medium-sized arteries throughout the body. Generally, renal arteries are the most affected with a proportion of 60%–70% of cases. Cervicocephalic arteries, including the carotid artery is then followed (25%–30%).[1,2] Some patients with carotid FMD may develop a stroke, which most commonly occurs in young patients without risk factors.[3]

FMD is classified into three histological variants, depending on the affected arterial wall.[4] FMD type 1, which affects the tunica media, accounts for 85% of the cases, and corresponds to areas of thin media with alternating fibromuscular ridges containing collagen, that replaces the normal tissue. FMD type 2 or intimal fibroplasia, accounts for 10% of the cases, showing circumferential or eccentric deposition of collagen within the tunica intima and the internal elastic lamina may be fragmented or duplicated. FMD type 3, which accounts for less than 5% of the cases, also known as periadventitial fibroplasia, are characterized by dense nodular deposits of collagen that replace the connective tissue of the adventitia.

Contrast angiography is one of the methods to diagnose FMD, where classical imaging is a "string of beads" pattern of the affected arteries caused by alternating areas of wall hypertrophy and narrowing. However, it is often difficult to definitively diagnose FMD based solely on angiographic findings[5]; the differentiation between FMD and vasculitis is especially difficult to make. A detailed histological examination of the three layers of the carotid artery is required for a definitive diagnosis of FMD. The high resolution of optical coherence tomography (OCT) allows easy and clear assessment of intravascular characteristics and morphology. In this case report, we describe the application of OCT during stent placement for FMD carotid artery stenosis. OCT for CAS is off-label use in Japan. This study was approved by the ethics committee of our institution (No. 00116).

[1]*Department of Neurosurgery, Shin-Komonji Hospital, Kitakyushu, Fukuoka, Japan*
[2]*Department of Neurosurgery, Fukuoka Wajiro Hospital, Fukuoka, Fukuoka, Japan*

Received: October 14, 2020; Accepted: October 21, 2020
Corresponding author: Shogo Oshikata. Department of Neurosurgery, Shin-Komonji Hospital, 2-5, Dairishinmachi, Moji-ku, Kitakyushu, Fukuoka 800-0057, Japan
Email: s.oshi1227@gmail.com

Case Presentation

A 30-year-old woman was diagnosed with mild left carotid artery stenosis and was diagnosed with a suspected case of FMD by MRA for the evaluation of chronic headaches. Carotid artery stenosis gradually progressed over the course of 7 years. MRA showed severe stenosis in the left cervical internal carotid artery (ICA) (**Fig. 1A**), and T1-weighted plaque image showed an iso-intensity signal (**Fig. 1B**). Carotid artery stenting (CAS) was therefore recommended. The patient did not have any risk factors for atherosclerosis such as a history of diabetes mellitus, hyperlipidemia, and hypertension. Moreover, possible presence of FMD was not seen in the renal and other cervicocephalic arteries. Laboratory examinations were negative for antinuclear antibody, matrix metalloproteinase-3, proteinase-3 antineutrophil cytoplasmic antibody (R3-ANCA), and myeloperoxidase ANCA.

Intervention

CAS was performed using a balloon-guided catheter and distal filter protection. Common carotid angiography revealed severe stenosis at a high position in the cervical ICA (**Fig. 2A**). Dragonfly (St. Jude Medical, St. Paul, MN, USA) OCT system was used. The procedure for OCT has been described previously.[6] Pre-procedural OCT revealed a thickened media in the distal portion of the stenosis (**Fig. 2B** and **2C**); most stenotic lesions also showed a thickened media, (**Fig. 2D** and **2E**). Non-stenotic lesions and thickened media were also observed proximally (**Fig. 2F**). Therefore, the lesion was diagnosed as FMD type 1 based on the OCT finding.

Pre-dilatation was performed with a 3.5-mm balloon and a Precise (Cordis, Miami Lakes, FL, USA) 7-40 mm stent was deployed. Stenotic lesion could not be fully dilated with the stent; therefore, an additional Precise 8 mm–40 mm stent was deployed with partial overlapping. Post-dilatation was performed with a 4.5-mm balloon. Post-procedural OCT showed excellent dilatation and good stent apposition (**Fig. 2G**) with a little tissue protrusion through the stent struts (**Fig. 2H**). A final angiography revealed an excellent dilatation of the lesion (**Fig. 2I**). Post-procedural course was uneventful. Ultrasonography performed 6 months after the procedure showed good stent patency.

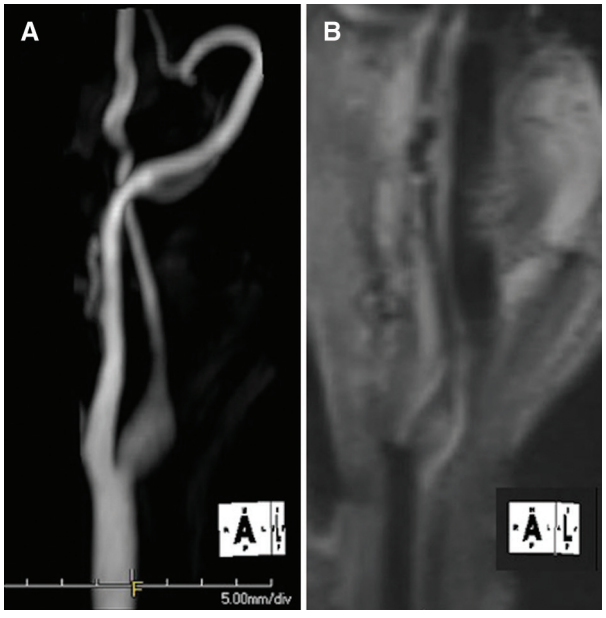

Fig. 1 (**A**) MRA showed severe stenosis in the left cervical. (**B**) MRI T1-weighted plaque image showed an iso-intensity signal.

Discussion

FMD is most often diagnosed clinically and by the characteristic appearance on angiography; however, the presence of a focal or long tubular stenotic pattern can lead to misdiagnosis.[5,7] Furthermore, it is difficult to differentiate diffuse intimal FMD from vasculitis. Without the availability of histological proof of FMD or inflammation, it may be difficult to distinguishing these entities, as their angiographic appearances may be similar. While the CAS procedure, typically, uses intravascular ultrasound (IVUS) to assess the lesions, IVUS has limitations in assessing the details of the vessel wall structures. On the other hand, the resolution of OCT (10–20 μm) is about 10 times that of IVUS (100–150 μm).[8] Therefore, OCT can distinguish between internal and external thin layers and is a better ability to evaluate the intravascular properties and morphology of FMD than IVUS. Although angiography was useful for confirming the diagnosis of FMD, OCT findings could be an adjunct tool if the diagnosis, degree of stenosis, and therapeutic effect remain uncertain.[9] Therefore, it is important to evaluate the detailed intravascular findings of causative and non-causative lesions in FMD using OCT. Since the use of OCT for CAS is not applicable in Japan in present, it is necessary to consult with the Ethics Committee in advance at each facility.

FMD is classified into three histological variants, depending on the affected arterial wall.[4] There are several reports of OCT findings of FMD from the renal and coronary arteries.[7,10,11]. And they reported that the abnormal segments showing many abnormalities not typically

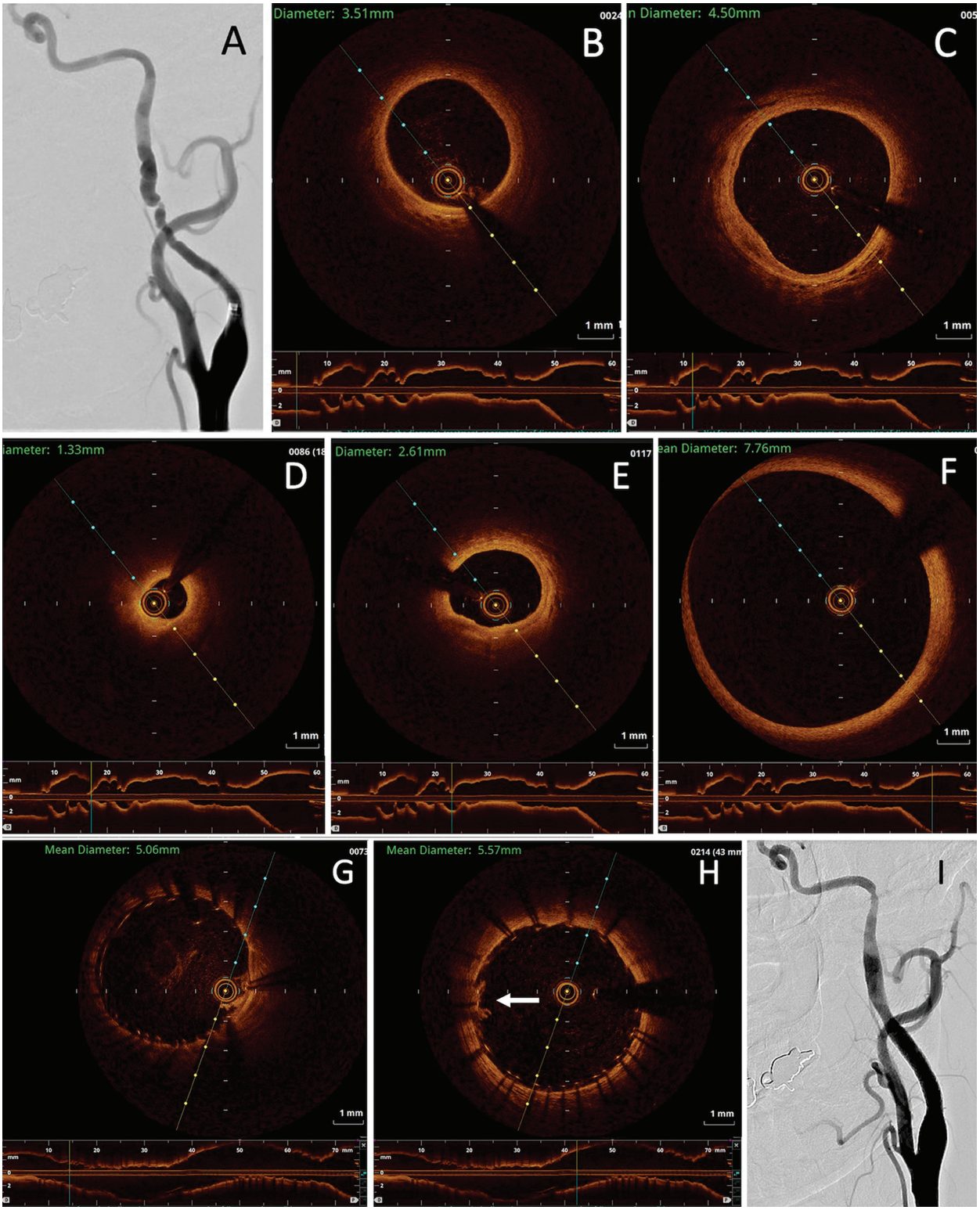

Fig. 2 (**A**) Pre-treatment angiogram showed severe stenosis in left carotid artery in a high carotid lesion. (**B**, **C**) OCT of the distal portion of the stenosis showed irregularly layered thinned highly backscattering intima (signal-rich) and a thickened low backscattering (signal-poor) area, suggesting thickened media. (**D**) OCT of the stenotic lesion also showed a thickened media. (**E**) OCT of the proximal portion of the stenosis showed highly thinned backscattering intima and layered highly and poorly mixed backscattering, suggesting the presence of collagen and smooth muscle cells in the media. (**F**) OCT of the carotid bifurcation showed thickened media, suggesting FMD was expanded below the common carotid artery. (**G**) After stent placement, good dilatation of the culprit lesion was obtained with good stenting apposition by OCT. (**H**) Small tissue protrusion was found in OCT (arrow). (**I**) Post-treatment angiogram showed excellent dilatation of the carotid artery. OCT: optical coherence tomography

observed with atherosclerosis, including multiple areas of patchy or diffuse intimal, medial, or adventitial abnormalities with thickening or accumulation of varied reflectivities, macrophage infiltration, loss or duplication of elastic membranes, and cavitation. In our case, pre-procedural OCT revealed thickened media in the whole carotid artery suggesting FMD type 1, and no findings suggestive of inflammation and atherosclerosis such as macrophage infiltration, lipid, and plaque disruption were seen. In addition, in this case, FMD was discovered by OCT at the distal ICA (at the level of the first cervical vertebra), up to the point accessible by OCT. In the proximal lesion, FMD was found to be at the level of the carotid bifurcation. In this case, FMD ranged at least from the carotid bifurcation to the cervical ICA. Proximal and distal edges of the FMD could not be determined because of the limitation of navigation of the OCT probe and the penetration depth of OCT (5–6 mm).[6] Although OCT has such limitations, OCT could help confirming the diagnosis of FMD and accurately determine the extent of the lesion. OCT was also useful to evaluate stent malapposition and tissue protrusion after post-stent placement. In this case, insufficient stent coverage after the first stent placement and a sufficient stent coverage after additional stenting were observed.

Conclusion

OCT evaluation during CAS allowed for a definitive diagnosis of FMD because the OCT images revealed a detailed finding of the three layers of the carotid artery.

Disclosure Statement

There is no conflict of interest for all authors regarding to this article.

References

1) Mettinger KL: Fibromuscular dysplasia and the brain. II. Current concept of the disease. *Stroke* 1982; 13: 53–58.
2) Leary MC, Finley A, Caplan LR: Cerebrovascular complications of fibromuscular dysplasia. *Curr Treat Options Cardiovasc Med* 2004; 6: 237–248.
3) Corrin LS, Sandok BA, Houser OW: Cerebral ischemic events in patients with carotid artery fibromuscular dysplasia. *Arch Neurol* 1981; 38: 616–618.
4) Stanley JC, Gewertz BL, Bove EL, et al: Arterial fibrodysplasia. Histopathologic character and current etiologic concepts. *Arch Surg* 1975; 110: 561–566.
5) Begelman SM, Olin JW: Fibromuscular dysplasia. *Curr Opin Rheumatol* 2000; 12: 41–47.
6) Harada K, Oshikata S, Kajihara M: Optical coherence tomography evaluation of tissue prolapse after carotid artery stenting using closed cell design stents for unstable plaque. *J Neurointerv Surg* 2018; 10: 229–234.
7) Mizutani K, Itoh A, Sugioka K, et al: Intravascular findings of fibromuscular dysplasia on optical coherence tomography. *J Cardiol Cases* 2015; 12: 39–42.
8) Sengottuvelu G, Rajendran R, Majumdar D: Optical coherence tomogram of spontaneous coronary artery dissection managed with drug eluting stent. *Indian Heart J* 2014; 66: 247–248.
9) Niizeki T, Ishino M, Kitahara T, et al: Endovascular therapy for fibromuscular dysplasia of the bilateral external iliac arteries visualized with optical coherence tomography. *Am J Case Rep* 2015; 16: 187–190.
10) Rajesh V, Kewal K, Darshan K, et al: Optical coherence tomography in varying aetiologies of renal artery stenosis: a case series. *Eur Heart J Case Rep* 2019; 3: 1–6.
11) Saw J, Bezerra H, Gornik HL, et al: Angiographic and intracoronary manifestations of coronary fibromuscular dysplasia. *Circulation* 2016; 133: 1548–1559.