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Case Report

Effective endovascular reconstruction using multiple leo stents and silk flow-diverting stents for asymptomatic extracranial internal carotid artery dissection: a case report

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ABSTRACT

Extracranial carotid artery dissection (ECAD), although infrequent, represents the predominant form of artery dissection within the brain. It accounts for 25% of young-onset ischemic strokes in adults. Its atypical presentation and limited availability of high-quality evidence underscore the importance of precision medicine in its management. This study aimed to illustrate a combined approach utilizing multiple scaffolding Leo stents and Silk flow-diverting stents to manage asymptomatic ECAD a week after the onset of acute ischemic stroke. A 40-year old Indian male with uncontrolled hypertension was admitted due to acute vestibular syndrome. His brain computed tomography (CT) scan showed cerebellar infarct. However, his angiographic cervical CT scan showed asymptomatic ECAD, which was confirmed with digital subtraction angiography. He was therefore managed with optimal antihypertensive agents and dual antiplatelet. The endovascular reconstruction procedure initiated one week after the onset of stroke by employing one scaffolding Leo stent and one Silk flow-diverting stent at his right internal carotid artery. He was discharged three days following the endovascular procedure and presented no complications until three-month follow-up. This safe approach could be considered for individuals with ECAD to help prevent secondary strokes, particularly among those in the productive age group.

Keywords: Acute ischemic stroke, Dissecting aneurysm, Endovascular reconstruction, Extracranial carotid artery dissection

INTRODUCTION

Cervicocephalic artery dissection is an uncommon etiology of ischemic stroke listed under "stroke of other determined etiology" according to the etiology classification from trial of Org 10172 in acute stroke treatment (TOAST).[1]

It is characterised by hematoma in the wall of cervical or intracranial arteries which was due to the tear in the arterial tunica intima, primary mural hematoma, or traumatic events including shear injury from extreme neck movement, direct laceration from surrounding structure, or direct vascular injury.

Extracranial cervical artery dissection (EAD), specifically extracranial carotid artery dissection (ECAD), is the most common type of cervicocephalic artery dissection. It is reported to be rare in general population, reaching 2.6-3 new symptomatic cases per 100.000 population annually. However, this number was predicted to be higher due to its possible asymptomatic variants.[2] It also serves as the etiology of up to 25% cases of acute ischemic stroke under 50 years old, which may reduce quality of life, productivity, and produce significant economic burden. Mortality was less than 5% and good recovery (modified Rankin scale 0-2) may be reached in 75-92% cases of ECAD, but functional recovery from its complication of stroke was similar to any other etiologies of stroke.[3]

Besides its well-known clinical manifestation of stroke/transient ischemic attack (>50%), challenges in the management of ECAD arises from its unrecognized atypical symptoms including headache and neck pain (65-95%), third order Horner's syndrome (25%), pulsatile tinnitus (8%), isolated to multiple cranial neuropathies (12%), and compressive cervical radiculopathy most commonly at C5-C6. These symptoms may be due to mass effect and local compression of adjacent structure, arterial *in situ* stenosis, or distal thromboembolism. Extracranial carotid artery dissection may also produce long-term complications including dissecting pseudoaneurysm (19%), recurrent stroke/TIA within first few weeks following the dissection (0-13%), and recurrent dissection within the first two months (<5% to 9.2%).[2,3]

In addition, its low prevalence may also explain the sparse clinical trial and guidelines regarding the recommended management of ECAD. European Stroke Organization recommended that intravenous thrombolysis and mechanical thrombectomy can be performed in ECAD with hyperacute stroke in accordance to their standard eligibility criteria, albeit with very low quality of evidence and very weak strength of recommendation. There is also no recommendation regarding the definitive management of endovascular or surgical treatment in either symptomatic or asymptomatic ECAD.[4,5] Precision medicine is therefore required to manage this rare but debilitating disease.

This case report aimed to present a case of acute ischemic stroke with asymptomatic ECAD who was successfully treated with the combination of endovascular treatment modalities using Leo self-expanding braided nickel-titanium alloy (nitinol) stent and Silk flow-diverting stents. This approach may expand the experience in managing ECAD especially in Indonesia.

CASE REPORT

A 40-year old male, Indian race, experienced sudden onset of vertigo a day prior to his admission. He also had uncontrolled hypertension. Physical examination showed an increased arterial blood pressure of 170/90 mmHg, regular pulse of 104 beats/minute, Glasgow coma scale of 15, and no lateralization.

Brain computerized tomography (CT) scan showed lacunar infarct at cerebellum. Additional ancillary diagnostics of brain angiography CT scan showed dissecting aneurysm at extracranial right internal carotid artery (RICA) (Figure 1). The right ECAD and dissecting aneurysm were also confirmed with digital subtraction angiography (DSA). The patient was managed symptomatically with blood pressure control. Secondary stroke prevention using dual antiplatelet was administered since the first day onset of stroke. The dissecting aneurysm at extracranial RICA was definitively treated using endovascular approach with the preparation of additional loading of aspirin 320 mg and clopidogrel 300 mg at the day of procedure. The procedure was performed at using the combination of a Leo self-expanding braided nitinol stent and a scaffolded Silk flow-diverting stent (Figure 2) from proximal C1 to C2 segment of RICA.

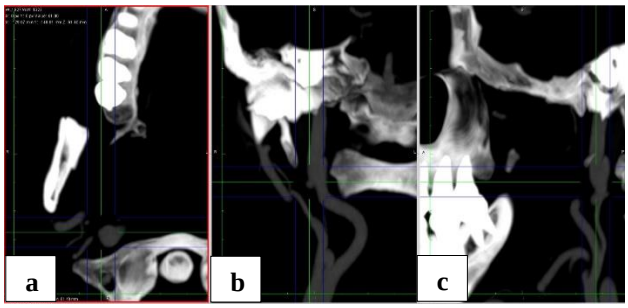

Figure 1: Brain angiography CT scan (a) the episode of admission using axial, (b) coronal, and (c) sagital view showed extracranial right internal carotid artery dissection with dissecting aneurysm.

Figure 2: Brain angiography CT scan with 3D reconstruction showed (a) extracranial right internal carotid artery dissection with dissecting aneurysm, and (b) which was managed using endovascular apporach with two Leo stents and two Silk flow-diverters.

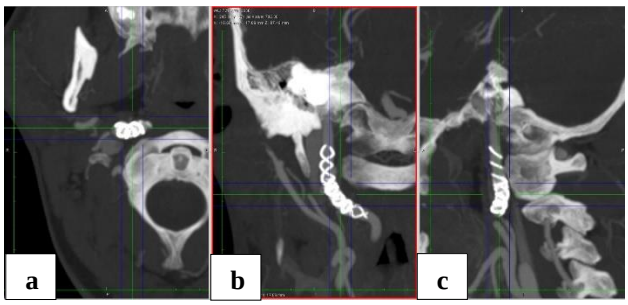

Figure 3: Brain angiography CT scan at six-month follow up using (a) axial, (b) coronal, and (c) sagital view showed successfully treated extracranial right internal carotid artery dissection with hyperdense stents and flow-diverters.

The patient was discharged three days following the procedure. Six-month follow up showed successful treatment with no ECAD (Figure 3). Double antiplatetet was resumed for a year.

DISCUSSION

Extracranial carotid artery dissection is a rare but significant etiology of stroke especially in young-to-middle age population. Its uncommon prevalence and atypical course of disease resulted in the low awareness and scarcity of evidence-based treatment recommendation for EAD. This case described a successful endovascular approach to EAD using two Leo stents and two Silk flow diverter performed in subacute phase of acute ischemic stroke for managing asymptomatic EAD.

This patient had sudden onset of vertigo which was consistent with lacunar infarct at the cerebellum from his CT scan. He was consequently diagnosed with an acute ischemic stroke of one-day onset. Hypertension has been largely recognized as the leading cause of ischemic stroke, including in this patient. Etiologic work-up using brain CT angiography showed no stenosis at his vertebral arteries, but there were EAD at his extracranial RICA. This extracranial RICA dissection was considered asymptomatic because this topical diagnosis was not directly responsible to the lacunar infarction at the cerebellum.

Extracranial cervical artery comprised five layers, including tunica intima or endothelium, internal elastic lamina, tunica media or muscular layer, external elastic lamina, and tunica adventitia from the lumen outward, respectively. Tunica intima and media provided structural and mechanical protection to vascular wall. Intracranial artery had little internal elastic lamina, no external elastic lamina, and little adventitial tissue which provided weaker support. The boundary between extracranial cervical and intracranial arteries was at the horizontal segment of the cavernous segment (C5) for internal carotid arteries and 0.5 cm after dural perforation (segment V4) for vertebral arteries.

Extracranial cervical artery dissection can manifest spontaneously or result from traumatic incidents. Spontaneous extracranial cervical artery dissection encompasses the more prevalent extracranial carotid artery dissection (ECAD) and the less frequent extracranial vertebral artery dissection (EVAD).[2]

Improvements in imaging technology have led to enhanced detection rates of spontaneous ECAD, revealing a prevalence of 1.2-3 cases per 100,000 individuals annually. However, this number may be underestimated due to the presence of asymptomatic ECAD, atypical symptoms ECAD, and the low medical resources in developing countries. Spontaneous ECAD tended to be male with an average age of 45 years whereas vertebral artery dissection tended to occur at intracranial segement.[2] The patient in this case was a 40-year old male, which was in conjunction with the demographic characteristics of ECAD in previous studies.

The etiology and pathophysiology of spontaneous ECAD were elusive, with current understanding suggested to be multifactorial. It occurred due to the interaction of the unmodifiable factors of age, genetics, and hormones with modifiable factors including inflammation and environmental stressors. Multiple risk factors that weakened or induced stress at the vascular wall included higher arterial curvature, connective tissue abnormalities, inflammation, and autoimmune diseases. Connective tissue abnormalites reported to be associated with ECAD included Marfan syndrome, Ehlers-Danlos syndrome, Turner's syndrome, and Loeys–Dietz syndrome. Inflammation due to infection or autoimmune diseases including systemic lupus erythemathosus, Sjogren syndrome, myasthenia gravis, autoimmune thyroid disease, and Takayasu arteritis may also predispose to ECAD by inducing the secretion of proinflammatory cytokines and promoting extracellular matrix protein degradation. Migraine was associated with twice increased risk to ECAD, which was thought due to the similar single nucleotide polymorphysm between migraine and ECAD.[2]

While being rarely reported, estrogen may explain the lower prevalence of ECAD in female. The involvement of inflammation and hormonal factors were supported by evidence showing improvement in spontaneous ECAD after the administration of high-dose hormones or anti-inflammatory medications. Postpartum period was also reported to increase the risk of spontaneous ECAD. In addition, vascular risk factors of hypertension, smoking, and increased homosystein level may predispose to spontaneous ECAD. The presence of uncontrolled hypertension in this case may predispose to the coincidence of acute ischemic stroke and spontaneous ECAD.[2]

The interaction of previously describe risk factors predisposed to two pathological conditions, which were vessel wall degeneration which weakened the vascular structure and mechanical wall stress. These conditions resulted in intima-media tear and/or hemorrhage in the vessel wall, the production of false lumen, narrowing of true vascular lumen, reduced blood flow, thromboembolism, and hypoperfusion which manifested with heterogenous symptoms. The immediate complications of ECAD included neck pain and headache (65-95%), nerve compression (e.g. third order post-ganglionic Horner syndrome (25%), isolated or multiple III, IV, V, VI, IX, X, XI, XII cranial nerve palsy (12%), pulsatile tinnitus (8%)), until retinal ischemia and ischemic stroke (more than 50% cases) whereas the long-term complication included pseudoaneurysm. The nonspecific nature of these symptoms may contribute to misdiagnosis and delay in the early management of ECAD until its gradual progression of neurological deterioration, which may occur even up to a month following the onset of ECAD.[2,3] For instance, the patient in this case did not report any nonspecific symptoms of ECAD, which was therefore determined as asymptomatic ECAD. The role of

imaging, in addition to the early symptoms awareness, is therefore paramount to early management of ECAD.

The characteristics of ECAD in imaging was double lumen, flame-shaped contrast-tapering enhancement (tapering stenosis), intimal flap, intramural hematoma, and dissecting aneurysm or pseudoaneurysm, of which the former two were the most common findings. Carotid duplex is a noninvasive screening tool which can measure flow dynamics, but requires high skill and had limitation in detecting ECAD higher than mandibule. Cervical angiography CT or MR may overcome that limitation, but CT had challenges in those with contrast allergies, kidney impairment, and contraindications of radiation including pregnancy and children whereas MRI needed a longer time to be performed. Digital subtraction angiography is the gold standard in diagnosing ECAD which is able to describe pathologies of vascular morphology and flow. However, it is more invasive, expensive, and poses risks including vascular perforation and contrast induced nephropathy.[2,3]

Angiographic CT was the recommended imaging modalities by American Heart Association to screen for ECAD due to its better spatial resolution especially for small-caliber arteries, its faster procedure, its more common availability, and its superiority to detect pseudoaneurysm and intimal flap, with sensitivity and specificity of 65% and 100% compared with DSA, respectively. However, angiographic MR with fat saturation was more superior in providing better detection for small intramural hematoma, better vessel wall resolution, and better detection for small ischemic stroke, with sensitivity and specificity of 50-100% and 30-100% compared with DSA, respectively.[3] The patient in this case had cervical angiography CT for routine vascular risk factor screening in stroke, which described an incidental finding of ECAD with dissecting aneurysm. This finding was also confirmed with DSA.

The primary aim of the management of ECAD was the prevention of ischemic complications. Spontaneous resolution of ECAD had been reported in some cases, but the protective prognostic factors were still unknown.[2,3] However, management for thromboembolism prevention and hemodynamic complication must be performed. Hyperacute ischemic stroke due to ECAD should be managed in a similar protocol to general hyperacute ischemic stroke, which included intravenous thrombolyis and mechanical thrombectomy, whereas acute ischemic stroke may be managed using anticoagulant or antiplatelet.[4] The patient in this case received antiplatelet for secondary stroke prevention due to its admission time beyond the time window for reperfusion management.

There were only very low evidence regarding the eligibility criteria, benefit vs risk, and ideal time for endovascular and surgical approach in ECAD. However, expert consensus from European Stroke Organization suggested that patients with ECAD and deterioration such as recurrent ischemic events despite optimal antithrombotics or expanding dissecting aneurysm causing compression may benefit from those approaches.[4] Our patient was a relatively young patient with ECAD which had dissecting aneursym that was therefore considered to have endovascular treatment performed.

Open repair was the gold standard management of ECAD. However, endovascular treatment, which was less invasive, had been increasing reported to be successful as well. Cornwall et al analyzed seven symptomatic and ten asymptomatic ECAD patients who had endovascular approach performed and reported no perioperative and postoperative complications after follow-up of 338 (8-3,039) days. The endovascular approach was suggested to be determined from the anatomy characteristics of ECAD, including the presence of internal carotid artery occlusion, the presence of arterial extravasation and rupture, and the size of adjacent vessel diameter (Figure 4).

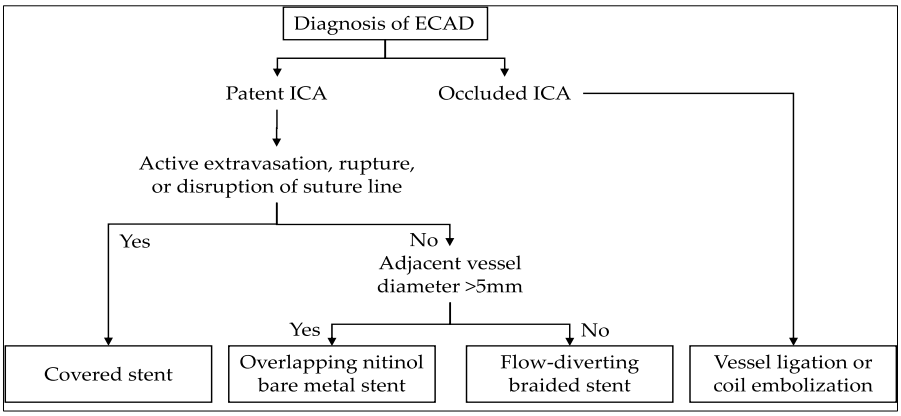

Figure 4: Suggested endovascular approach to extracranial carotid artery dissection.

Our patient was treated with combined strategy of one Leo self-expanding braided nitinol stent and one Silk flow-diverting stent. Overlapping nitinol stents were applied due to the absence of active extravasation with the aim to support the dissecting vessel wall. Although no single stent stood out as superior to the others, when compared to the Enterprise and Solitaire stents, the Leo stent emerged as distinctive. Unlike its counterparts, Leo is uniquely crafted

from a single self-expanding nitinol wire, boasting superior radial strength for oversizing ranging from 15% to 50%. It exhibits low bending stiffness, exceptional kink resistance, minimal ovalization, extensive bending wall coverage, and the smallest cell size. Additionally, it offers the advantage of repositioning after partial deployment. These characteristics endow Leo with robust capabilities for vascular reconstruction and vessel wall protection. While primarily indicated for intracranial aneurysms, Leo stent has also been documented in a few case reports of vessel bifurcation disease, alongside the Enterprise and Solitaire stents.[6] In addition to Leo stents, Silk flow diverting stent was applied therafter to ensure the vessel patency and flow. Silk was composed with 48 nitonol-braided microfilaments and four platinum wires, flared end, with metal coverage of 35-55% and retrievable. Nitinol had superelastic and shape-memory properties whereas the use of many braided microfilaments reduced the shortening rate of the flow diverter upon full expansion and provides more suitable pore size modulation.[7,8] The combination of Leo stent and Silk flow diverting stent provided strong reconstruction and maintaned normal flow to the respective internal carotid artery.

The best timing for endovascular management of ECAD was still unknown as well.[4,5] Therefore, the decision of different types of endovascular approach and the timing of ECAD reconstruction is still tailored-made. We conducted the procedure one week after the onset of ischemic stroke because the ECAD was asymptomatic and the patient had already passed beyond the acute phase of the stroke.

The annual mortality rate of ECAD even under long-term medical therapy was been reported to be 8–12%. Endovascular management from 201 subjects by Xianjun et al reported a technical success rate of 99.1%, overall periprocedural major cardiovascular event rate of 4%, and no periprocedural mortalities. Post-procedural complication following endovascular management included intimal hyperplasia, in-stent restenosis, or occlusion of the treated vessel in 3.3% subjects and recurrent transient ischemic attacks in the respective vessel in 2.1% subjects after mean follow-up duration of 20.9 months. Our patient had been followed-up three months post-procedure and had no reported complications. Therefore, it was safe to perform endovascular procedure using combined Leo stents and Silk flow-diverting stents in asymptomatic ECAD following acute phase of stroke as secondary stroke prevention.

CONCLUSION

Extracranial carotid artery dissection is uncommon, yet it accounts for 25% of young-onset strokes. Its varied and often atypical presentation may lead to delays in diagnosis and treatment. Currently, guidelines on optimal management strategies for ECAD are limited and its evidence quality is low. Therefore, precision-medicine approach involving multidisciplinary team is crucial for effective management. Alongside traditional treatments like antiplatelet or anticoagulant therapy, endovascular interventions offer a promising minimally invasive option for addressing ECAD even in asymptomatic subjects. Combined multiple Leo and Silk stents were a possible alternative of endovascular approach to manage ECAD. The goal is to prevent future strokes, particularly in individuals of productive age.

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