



Case Report

Recurrence of internal carotid artery dissection associated with elongated styloid process: A case report

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ABSTRACT

Background: An elongated styloid process is known to cause ischemic stroke. Previous reports claim that internal carotid artery (ICA) dissection due to the elongated styloid process has good outcomes when treated conservatively; however, long-term follow-up has not been attempted and recurrence in the later period has not been reported so far. We report a case of recurrence of symptoms over a decade after the initial onset.

Case Description: A 59-year-old man experienced a transient ischemic attack (TIA) 10 years ago. Six years ago, he experienced hemispheric TIA, and magnetic resonance angiography revealed a diminished signal of the left ICA; however, no further examination was performed. Four years ago, he experienced another transient amaurosis attack and was treated with antiplatelet therapy because no embolic source was detected using ultrasonography examination, and he was diagnosed with idiopathic ICA dissection. Recently, he experienced a third amaurosis fugax attack. Digital subtraction angiography and cone-beam computed tomography demonstrated left cervical ICA dissection due to elongated styloid process. He underwent surgical resection of the left styloid process and cervical stent placement. He had no ischemic attacks postoperatively.

Conclusion: The elongated styloid process may cause recurrent ischemic attacks over a decade due to ICA dissection.

Keywords: Elongated styloid process, Internal carotid artery dissection, Recurrence

INTRODUCTION

The elongated styloid process is one of the causative diseases of stroke, where ischemic neurologic symptoms are caused by the compression of the cervical internal carotid artery (ICA) because of the elongated styloid process or calcification of the stylohyoid ligament. ICA dissection or compression due to elongated styloid process is a rare condition; therefore, it is often misdiagnosed as idiopathic ICA dissection.

Most with ICA dissection due to elongated styloid process have been treated conservatively and have been reported to have a good course;^[1,10,15-17,19,21] however, there are no long-term follow-up reports, and the long-term prognosis is unknown.

We present a case of cervical ICA dissection due to elongated styloid process which recurred over a long period and repeatedly showed symptoms. We also review articles on the management

of these uncommon lesions, particularly to the recurrence or deterioration of the lesions after conservative or endovascular treatment.

CASE DESCRIPTION

A 59-year-old man with a history of medically treated hypertension and hyperlipidemia had a transient ischemic attack (TIA) and was transferred to another hospital 10 years ago; however, the details about the case were unknown. Six years ago, he had a left transient amaurosis attack and magnetic resonance angiography (MRA) revealed a decreased signal of the left ICA [Figure 1a]; however, no detailed examination was performed. Four years ago, he experienced another transient amaurosis attack on the left side and visited our hospital. Although MRA showed a further decreased signal of the ICA [Figure 1b], conservative therapy with an antiplatelet was delivered for a few months since an embolic source was not detected at the origin of the ICA using carotid duplex ultrasonography examination, and he was diagnosed with idiopathic cervical ICA.

Recently, the left amaurosis fugax reappeared, and the patient was referred to our hospital. Oral administration of clopidogrel (75 mg/day) and cilostazol tablets (100 mg/day) managed the amaurosis fugax. Due to repeated amaurosis attacks, with the ophthalmic assessments to exclude other ocular or retinal pathology, and gradually decreasing left ICA on MRA [Figure 1c], we performed computed tomography angiography (CTA) and digital subtraction angiography (DSA). CTA

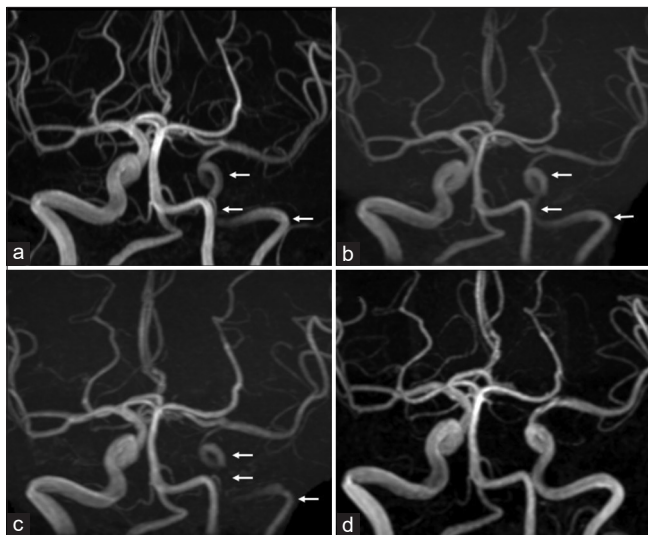


Figure 1: Magnetic resonance angiography (MRA) images of the patient. MRA obtained 6 years ago (a), 4 years ago (b), and preoperatively (c) demonstrating that the signal of the intracranial left internal carotid artery (ICA) is gradually reduced (white arrow). MRA obtained after the resection of styloid process and carotid artery stenting (CAS), demonstrated the improvement of the ICA signal (d).

demonstrated that the left elongated styloid process was directly impinged on the ICA [Figure 2a]. The DSA and cone-beam computed tomography (CT) demonstrated that the bilateral styloid process was elongated, and the left cervical ICA had an irregular lumen and stenosis with false lumen [Figure 2b and c]. Because of proximity between the ICA stenosis site and tip of the elongated styloid process, the patient was finally diagnosed with repeat left cervical ICA dissection caused by the compression of the left elongated styloid process.

The patient underwent left styloid resection through transcervical approach. In the surgery, we confirmed that the tip of the styloid process was very close to the ICA [Figure 3a], and we removed the 2 cm tip of the styloid process [Figure 3b and c]. Post the removal of the styloid process, no contact was verified on the image study between the styloid process and cervical ICA [Figure 2d].

In addition, the patient underwent stent placement (Stent: Precise 6 × 20) since we considered that the residual severe stenosis caused by the left cervical ICA dissection could lead to further ischemic events [Figure 2e and f]. He was discharged without any complications associated with surgery. Postoperatively, he experienced no ischemic attack, and the MRA 3 months after these procedures showed improvement of the left MRA signal [Figure 1d].

DISCUSSION

We present a case of recurrent ischemic symptoms after 10 years caused by cervical ICA dissection due to elongated styloid process, although without an accurate diagnosis. To the best of our knowledge, 15 cases of progression or recurrence after initial therapy due to elongated styloid process have been described so far [Table 1]. Some clinical suggestions from our case are as below.

First, clinicians must distinguish between “idiopathic” ICA dissection and “traumatic” ICA dissection caused by the elongated styloid process. Otherwise, preventable embolic stroke may reoccur. Several reported cases of ICA dissection with elongated styloid process have recurred or worsened even after diagnosis and administration of oral antithrombotic agents [Table 1]. The recurrence of ischemic events or progression of the dissection are rare in idiopathic cervical ICA dissection, and the recurrence rate in the same vessel after remission was reported to be 0–0.0135%.^[2,3,11,14] ICA dissection due to the elongated styloid process was determined to have higher recurrence risk than that of idiopathic ICA dissection.^[7,18] Conversely, the possibility of elongated styloid process when ischemic event recurs should be considered. Moreover, some cases of ischemic event recurrence have led us to identify the underlying etiology of elongated styloid process.^[4-6,8,20]

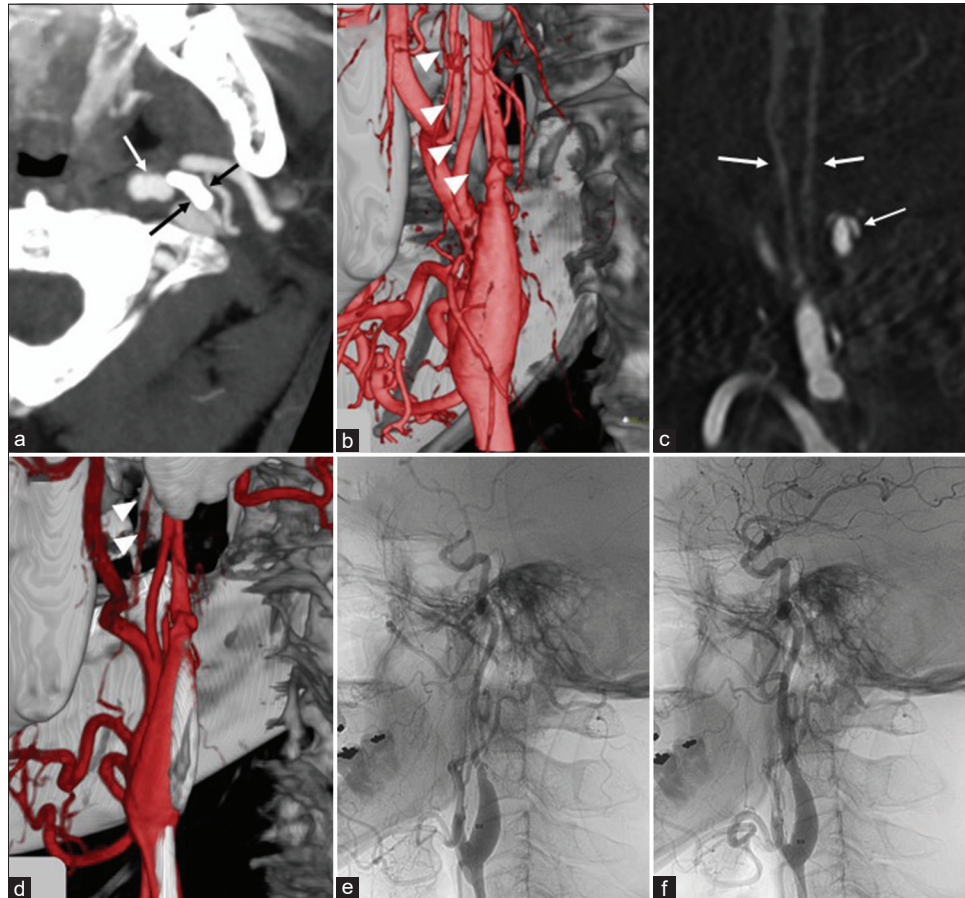


Figure 2: CT angiography, cone-beam computed tomography (CT), and digital subtraction angiography (DSA) of the patient. Preoperative CT angiography, axial image, demonstrating that left elongated styloid process (black paired arrows) is directly impinging on the ICA (white arrow) (a). Preoperative fusion image (cone-beam CT and 3D-DSA) demonstrating the tapered and irregular stenosis of the left internal carotid artery (ICA). The tip of elongated styloid process was very close to the ICA (arrowheads), and the ICA showed severe luminal stenosis (b). Preoperative 3D-DSA (CT-like images), sagittal view, demonstrating the false lumen (white arrow) associated with the elongated styloid process (white paired arrows) (c). Fusion image (cone-beam CT and 3D-DSA) obtained after the resection of elongated styloid process demonstrated the shortening of the styloid process (arrowhead), and the tip was no longer close to the ICA (d). The ICA angiogram (lateral view) before (e) and after (f) the procedure of carotid artery stenting (CAS) demonstrated the resolution of the left ICA stenosis.

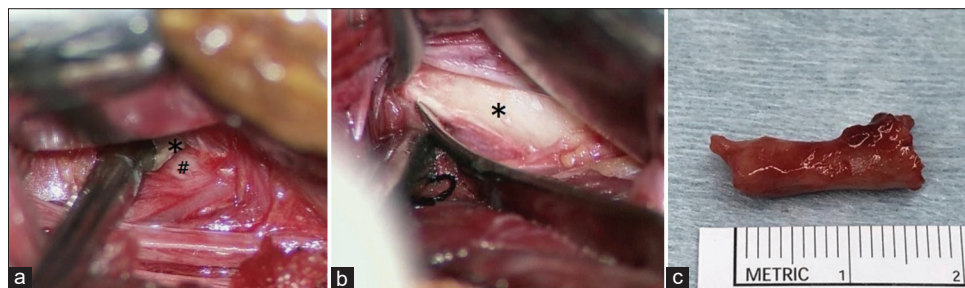


Figure 3: Intraoperative photographs during resection of the elongated styloid process. Using the infradigastric muscle approach, the tip of the styloid process (*) and cervical ICA (#) is in contact with each other (a). Using the supradigastric approach, the bony structure was stripped of from its periosteum and muscle (b), and the tip that is approximately 2 cm of the process was amputated and removed (c).

Cervical ICA dissection due to the elongated styloid process can cause recurrent ischemic events in the long term, and

long-term follow-up may be necessary, even if conservative treatment does not cause a short-term recurrence. Although

Table 1: Previous reports of internal carotid artery dissection caused by elongated styloid process that demonstrates symptom recurrence or image deterioration.

Age/sex	Side	Initial treatment	Exacerbation period*	Exacerbation event, reason	Treatment after recurrence	Postoperative follow-up period	Author	Year
57/M	Bil.	AG	1 day	ICO (Lt.)	ET, CAS	1 year	Todo T	2012
38/M	Lt.	ET (M1), CAS	4 months	Intrastent thrombosis	ET(M1), CAS (additional), RSP	6 months	Sveinsson O	2013
55/M	Bil.	AG	4 days	ICO (Lt.)	RSP (Bil.), CAS (Rt.)	3 months	Ogura T	2014
55/M	Bil.	AG	NA	ICO (Lt.)	RSP (Bil.), CAS (Rt.)	3 months	Naito Y	2014
41/M	Rt.	AG	5 months	ICO	ET (ICA terminus), CAS, RSP	12 months	Miyata	2016
64/M	Lt.	CAS	1 year	ICO, Stent fracture	None (as asymptomatic)	NA	Hooker	2016
49/M	Bil.*	None	3 years	Contralateral ICA dissection	RSP (Rt.), CAS (Rt.)	NA	Dewan	2016
47/F	Rt.	CAS	NA	TIA	RSP	NA	Subedi	2016
60/M	Rt.	AG	1 day	New infarct	CAS	3 weeks	Smoot TW	2017
39/W	Rt.	AG	3 months	Image deterioration	CAS	5 years	Mann	2017
48/M	Rt.	ET (M2), CAS	6 months	Stent displacement	CAS (additional)	21 months	Shimozato	2018
45/M	Lt.	None	5 weeks	Aneurysm formation	CAS, Coil embolization	4 months		
46/M	Bil.	CAS (Lt.)/AG (Rt.)	4 days	Image deterioration (Rt.)	CAS (Rt.), RSP (Bil.)	3 years	Torikoshi	2019
58/M	Lt.	ET (M2), CAS	3 months	Stent fracture, aneurysm formation	Coil embolization [#] , SP fracture	1 years	Yano	2019
46/F	Bil.	CAS (Bil.)	5 day	ICO, Intrastent thrombosis (Rt.)	RSP (Lt.)	NA	Horio Y	2020
59/M	Lt.	AG	4 years	TIA, Image deterioration	CAS, RSP	3 months	Present case	

AG: Antithrombotic agents, Bil.: Bilateral, CAS: Carotid artery stenting, ET: Endovascular thrombectomy, Lt.: Left, M1: Sphenoidal segment of the middle cerebral artery, M2: Insular segment of the middle cerebral artery, NA: Not available, RSP: Resection of styloid process, Rt.: Right, TIA: Transient ischemic attacks, *: Interval from first event or previous treatment to recurrence of symptom or deterioration on imaging, †: First pathogenesis is Lt. ICO due to ICA dissection, ‡: Stent-assisted coil embolization

most cases of ICA dissection recurrence due to elongated styloid process occur in the acute phase [Table 1], our patient exhibited symptoms over a long period even in the remote phase of dissection.

Many reports have indicated that conservative treatments such as antithrombotic therapy, with or without cervical rest, are effective.^[15] However, we consider that it is impossible to lead a normal life with a resting neck, and recurrence is unavoidable over a long period. Our intraoperative findings show the proximity of the styloid process tip and ICA and indicate that they can come in contact by mild cervical movement. The longer the styloid process is and the closer it is to the ICA, the higher is the risk of ICA dissection.^[9,12,13] In addition, cervical ICA was reported to move from the front to the back by 19.8 mm when the head was rotated to the left or right,^[6] although we could not demonstrate occurrence of this movement in our case.

Second, considering the risk of recurrence, it may be justified to consider surgical treatment aggressively – especially for

elongated styloid process resection – when diagnosing a patient with ICA dissection associated with the elongated styloid process. Most acute treatments for the progression have been performed by carotid artery stenting (CAS); however, the few complications followed by CAS may be due to the stent being affected by elongated styloid process such as stent fracture, intrastent thrombosis, and stent displacement [Table 1]. Therefore, it would be beneficial to perform styloid process resection before CAS for the treatment of stroke due to elongated styloid process, when possible. Another reason, we consider, to remove styloid process first before CAS is the requirement for antiplatelet agents after CAS which may increase risk of bleeding with surgery.

CONCLUSION

Clinicians should take into consideration the possibility of elongated styloid process when diagnosing spontaneous cervical ICA dissection to prevent the occurrence of future ischemic events.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Conflicts of interest

There are no conflicts of interest.

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