Case Report

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

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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; 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 amurosis 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 amurosis 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 amurosis 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 amurosis fugax. Due to repeated amurosis 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 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]
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...
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. 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. 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, 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.

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**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.) Intrastent thrombosis | ET, CAS                   | 1 year                        | Todo T | 2012 |
| 38/M    | Lt.  | ET (M1), CAS     | 4 months             | ICO (Lt.)                  | 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) RSP (Rt.), CAS (Rt.) | 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                        | CAS                        | 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 (additional)          | 5 years                      | Mann | 2017 |
| 48/M    | Rt.  | ET (M2), CAS     | 6 months             | Stent displacement         | CAS (Rt.), CAS (Bil.)     | 21 months                     | Shimozato | 2018 |
| 45/M    | Lt.  | None             | 5 weeks              | Aneurysm formation         | CAS, Coil embolization     | 4 months                      | Torikoshi | 2019 |
| 46/M    | Bil. | CAS (Lt.)/AG (Rt.) | 4 days             | Image deterioration (Rt.)  | CAS (Rt.), CAS (Bil.)     | 3 years                      | Yano | 2019 |
| 58/M    | Lt.  | ET (M2), CAS     | 3 months             | Stent fracture, aneurysm formation (Rt.) | Coil embolization*, SP fracture | 1 years                      | Horio Y | 2020 |
| 46/F    | Bil. | CAS (Bil.)       | 5 day                | ICO, Intrastent thrombosis (Rt.) | RSP (Lt.) | NA | Present case |
| 59/M    | Lt.  | AG               | 4 years              | TIA, Image deterioration   | CAS, RSP                   | 3 months                      | Present case | 2019 |

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
Declarations of patient consent
Institutional Review Board (IRB) permission obtained for the study.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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How to cite this article: Yokoya S, Takezawa H, Oka H, Hino A. Recurrence of internal carotid artery dissection associated with elongated styloid process: A case report. Surg Neurol Int 2021;12:473.