Left Mandibular Pain in a Patient of Aortic Dissection Presenting with Acute Ischemic Stroke: a Case Report

Chien-Hsin Li, Yung-Chu Hsu, Yung-Chu Hsu

Abstract-

Purpose: If a patient with aortic dissection (AoD) is inadvertently treated with thrombolytic agents, severe complications and poor outcomes are likely to ensue. We reported a case and postulated mandibular pain as warning sign for diagnosing AoD related stroke.

Case Report: We report an 81-year-old woman presenting with left mandibular pain followed by left hemiplegia. Extracranial carotid duplex showed dissection of the right common carotid artery and chest CT angiography proved AoD. She did not have any typical chest or back pain.

Conclusion: AoD should be carefully considered in patients with acute ischemic stroke presenting with mandibular pain. A high clinical alert and urgent CT angiography may help identify AoD.

Key Words: aortic dissection, common carotid artery dissection, stroke, carotid duplex, thrombolysis

Acta Neurol Taiwan 2014;23:129-133

INTRODUCTION

Acute aortic dissection (AoD) is a potentially life-threatening disease that 21% patients died before admission[1]. The immediate mortality rate in AoD is extremely high as 1% per hour during the first 24 hours, which calls for early diagnosis and intervention[2]. Patients with AoD typically present with sudden sharp chest or back pain. However, 5% to 15% patients are painless[3, 4], which make the diagnosis more difficult. In the previous study, thirty patients with AoD initially complained of neurological symptoms and only two-thirds of them reported chest pain[5]. Stroke occurred in 6% type A AoD that might be missed in patients without chest pain[6]. Early recognition of AoD in the stroke patients who are otherwise thrombolysis candidates is particularly challenging. We describe a patient presenting with a clinical picture of acute ischemic stroke who was eligible

From the Division of Neurology, Department of Internal Medicine, Ditmanson Medical Foundation Chia-Yi Christian Hospital, Chiayi City, Taiwan; Division of Neurology, Department of Internal Medicine, Ditmanson Medical Foundation Chia-Yi Christian Hospital, Chiayi City, Taiwan; Division of Neurology, Department of Internal Medicine, Ditmanson Medical Foundation Chia-Yi Christian Hospital, 539 Jhongsiao Rd., Chiayi City, Taiwan.

Received April 29, 2014. Revised August 1, 2014.
Accepted October 14, 2014.

Correspondence to: Chien-Hsin Li, MD, Division of Neurology, Department of Internal Medicine, Ditmanson Medical Foundation Chia-Yi Christian Hospital, Chiayi City, Taiwan.
E-mail: creatoreuk@yahoo.com.tw

for thrombolytic therapy but her underlying AoD was confirmed later.

CASE REPORT

An 81-year-old woman presented with sudden onset of severe left mandibular pricking pain, rapidly followed by left hemiplegia, gaze preference to the right, and slurred speech few minutes later. She arrived at our emergency department one hour after the onset. Her consciousness remained clear. She did not have pain in the chest, back, or neck. Her vital signs were normal, showing a blood pressure of 127/68 mmHg with a heart rate of 81 beats per minute. Her neurological deficits improved spontaneously except for mild left hemiparesis on arrival at our emergency department. Her left mandibular pain also partially improved.

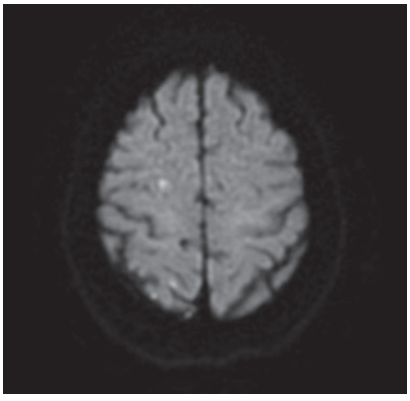

Figure 1. Multiple hyperintensities in the territory of right middle cerebral artery indicate acute ischemic stroke.

Her initial National Institutes of Health Stroke Scale score was 2. An electrocardiogram revealed normal sinus rhythm and a chest X-ray showed no evidence of widened mediastinum. The brain computed tomography (CT) was unremarkable. The cardiac enzymes were within normal limits. Intravenous thrombolytic treatment was withheld because of her rapidly improving neurological symptoms. Diffusion-weighted magnetic resonance imaging on day 1 showed multiple small acute ischemic lesions in the right hemisphere (Fig. 1). Extracranial carotid duplex disclosed a dissecting intima in the right common carotid artery (CCA) extending from the proximal part to the carotid bifurcation (Fig. 2A). Thus we further arranged CT angiography to survey the extent of artery dissection. Neck CT angiography demonstrated dissection of the right CCA and the aortic arch (Fig. 2B). Thoracic and abdominal CT angiography showed extension of the dissection to the abdominal aorta (Fig. 2C). The dissection was repaired by graft replacement of the ascending aorta and the aortic arch and open stent-grafting of the aortic arch and the descending aorta. Her left mandibular pain subsided after operation and neurological condition remained stable three months postoperatively.

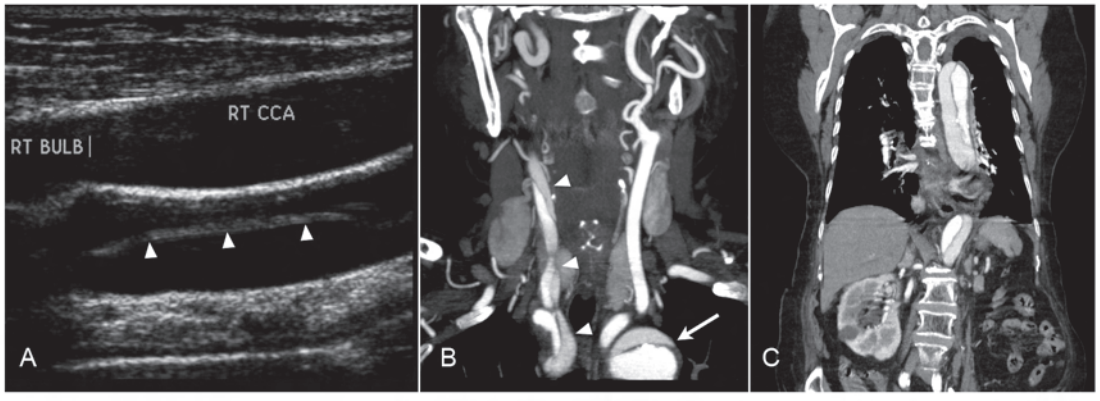

Figure 2. (A) Extra-cranial carotid duplex in longitudinal section showing a dissecting intima flap (arrowheads) in the right common carotid artery (CCA). (B) Neck CT angiography in the coronal plane demonstrating true and false lumens in the right CCA (arrowheads) and the aortic arch (arrow). (C) Chest and abdominal CT in the coronal plane showing involvement of the descending aorta.

DISCUSSION

We report a stroke patient with unusual prominent left mandibular pain, and carotid duplex showed right CCA dissection, which was extended from aortic arch confirmed by CT angiography. Her left mandibular pain was transient and temporally related to stroke. Right CCA dissection was unable to explain this pain since cervical artery dissection has been assumed to cause the ipsilateral pain. Dysethesia due to sensory cortical ischemia was less possible because: 1) her pain limited so precisely in the territory of left trigeminal mandibular branch, and 2) the lesion in brain magnetic resonance imaging (Fig. 1) was not compatible with left mandibular pain according to the anatomy of the homunculus. In clinical practice, left mandibular pain may be a referred pain of cardiac ischemia[7], but we excluded this possibility by cardiac examination. As we know, there is no literature discussing mandibular pain in patients of stroke caused by AoD, According to the anatomy, the thoracic cardiac branch of the vagus nerve carries visceral afferent input from the heart and great vessels. Research in rodents and primates found that noxious electrical and chemical stimulation of the thoracic branch of the left vagus nerve caused activation of left spinothalamic tract cells at the level of the trigeminothalamic tract. This provides a possible physiologic basis for the referred pain from heart and thoracic aorta to the trigeminal region[8], which could offer a better explanation of our patient's mandibular pain.

In the era of thrombolysis, clinicians have to select patients who are eligible for thrombolytic therapy within a time limit. If patients present stroke syndrome with concurrent headache or nuchal pain, it is worthy to evaluate the pain carefully. Cervical artery dissection, cerebral venous thrombosis, reversible cerebral vasoconstriction syndrome, cervical spinal epidural/subdural hemorrhage, and even aneurysmal sentinel headache should be listed in the differential diagnoses. When examining stroke patients with left mandibular pain, both myocardial infarction and AoD should be considered. The former is suitable for thrombolysis or heparinization therapy, but the latter is not. Although AoD is not a common cause of cerebral infarction, it may present as acute ischemic stroke in 5-10% patients[9,10]. Severe complications and poor outcomes are likely to ensue if a patient with AoD is inadvertently treated with thrombolytic agents because of the risk of fatal intra-cerebral hemorrhage, aortic rupture, and cardiac tamponade[11]. The most common site of pain caused by AoD is the chest, followed by the back and the abdomen[12]. The site of pain often indicates where the dissection starts. Anterior chest pain occurs in ascending aortic dissection and interscapular pain happens in descending aortic dissection. Neck and mandibular pain usually indicate dissection involving the carotid arteries and aortic arch, respectively[12]. Bossone et al have revealed arch vessel involvement was frequently found among the patients of AoD with stroke. As chest pain is less common in patients of AoD with stroke than those without stroke[6], clinicians must be aware of not only chest pain but also pain mentioned above in stroke patients.

In addition to clinical history, physical examination and chest radiograph are essential since some patients may be unconscious or aphasic. The classic findings of physical examination in patients of AoD are aortic regurgitation murmur and pulse deficit which were only noticed in 31.6% and 15.1% of patients, respectively[3]. Although up to 20% of chest x-rays may be negative, as in our presented case, it is a relatively simple and quick study that is an essential part before administration of rt-PA. Clinical suspicion of AoD should be aroused by the detection of a widened mediastinum on plain chest x-ray.

Unlike vertebral or internal carotid artery dissection, dissection of the CCA is rare but is associated with AoD in about one fourth of the cases. The other causes of CCA dissection included spontaneous, traumatic, and iatrogenic[13]. Dissection of the CCA, especially those on the right side, should raise the suspicion of AoD[14], as shown in our case. Acute ischemic stroke is caused by AoD through extension of the dissection into the CCA, thromboembolism, or cerebral hypoperfusion[3]. Duplex ultrasonography is a noninvasive diagnostic tool, and the key findings of dissection are a double lumen, mural thrombus, intraluminal hyperechoic/isoechoic lesion, and intimal flap[13]. As previous report[15], our detection of CCA dissection on extracranial carotid duplex aroused the suspicion of AoD and subsequent investigation of chest CT angiography confirmed the diagnosis of AoD, and timely avoided the catastrophic hemorrhagic complication of AoD which might have happened if thrombolysis or

anticoagulant therapy had been administered. However, emergent carotid duplex, is not often accessible in emergency department. CT angiography is a practical alternative choice in saving time to detect AoD before thrombolytic therapy. Although thrombolysis appears to be safe in patients with acute ischemic stroke due to cervical artery dissection[16], it is potentially harmful in those with AoD[17,18]. According to the guidelines for the diagnosis and management of thoracic aortic disease analyzing common twelve risk features as initial presentation of AoD[19], acute ischemic stroke in conjunction with pain is one of the high risk features that should be alert to.

In conclusion, AoD should be carefully considered in patients with acute ischemic stroke presenting with abrupt mandibular pain. A high clinical alert and urgent CT angiography may help detect AoD, which should be excluded from thrombolytic therapy.

REFERENCES

1. Meszaros I, Morocz J, Szlavi J, Schmidt J, Tornoci L, Nagy L, Szep L. Epidemiology and clinicopathology of aortic dissection. Chest 2000;117:1271-1278.
2. Braverman AC. Acute aortic dissection: clinician update. Circulation 2010;122:184-188.
3. Hagan PG, Nienaber CA, Isselbacher EM, Bruckman D, Karavite DJ, Russman PL, Evangelista A, Fattori R, Suzuki T, Oh JK, Moore AG, Malouf JF, Pape LA, Gaca C, Sechtem U, Lenferink S, Deutsch HJ, Diedrichs H, Marcos y Robles J, Llovet A, Gilon D, Das SK, Armstrong WF, Deeb GM, Eagle KA. The International Registry of Acute Aortic Dissection (IRAD): new insights into an old disease. JAMA 2000; 283:897-903.
4. Hsu YC, Lin CCK. Paraparesis as the major initial presentation of aortic dissection: report of four cases. Acta Neurol Taiwan 2004;13:192-197.
5. Gaul C, Dietrich W, Friedrich I, Sirch J, Erbguth FJ. Neurological symptoms in type A aortic dissections. Stroke 2007;38:292-297.
6. Bossone E, Corteville DC, Harris KM, Suzuki T, Fattori R, Hutchison S, Ehrlich MP, Pyeritz RE, Steg PG, Greason K, Evangelista A, Kline-Rogers E, Montgomery DG, Isselbacher EM, Nienaber CA, Eagle KA. Stroke and outcomes in patients with acute type A aortic dissection. Circulation 2013;128:S175-S179.
7. Kreiner M, Okeson JP, Michelis V, Lujambio M, Isberg A. Craniofacial pain as the sole symptom of cardiac ischemia: a prospective multicenter study. J Am Dent Assoc 2007;138:74-79.
8. Myers DE. Vagus nerve pain referred to the craniofa cial region. A case report and literature review with implications for referred cardiac pain. Br Dent J 2008; 204:187-189.
9. Khan IA, Nair CK. Clinical, diagnostic, and management perspectives of aortic dissection. Chest 2002;122: 311-328.
10. Iguchi Y, Kimura K, Sakai K, Matsumoto N, Aoki J, Yamashita S, Shibazaki K. Hyper-acute stroke patients associated with aortic dissection. Intern Med. 2010;49:543-547.
11. Fessler AJ, Alberts MJ. Stroke treatment with tissue plasminogen activator in the setting of aortic dissection. Neurology 2000;54:1010.
12. Nusair M, Abuzetun JY, Khaja A, Dohrmann M. A case of aortic dissection in a cocaine abuser: a case report and review of literature. Cases J 2008;1:369.
13. Zach V, Zhovtis S, Kirchoff-Torres KF, Weinberger JM. Common carotid artery dissection: a case report and review of the literature. J Stroke Cerebrovasc Dis 2012;21:52-60.
14. Uchino K, Estrera A, Calleja S, Alexandrov AV, Garami Z. Aortic dissection presenting as an acute ischemic stroke for thrombolysis. J Neuroimaging 2005;15:281-283.
15. Tsivgoulis G, Vadikolias K, Heliopoulos I, Patousi A, Iordanidis A, Souftas V, Piperidou C. Aortic arch dissection causing acute cerebral ischemia: an uncommon contraindication for intravenous thrombolysis. Circulation 2011;124:657-658.
16. Zinkstok SM, Vergouwen MD, Engelter ST, Lyrer PA, Bonati LH, Arnold M, Mattle HP, Fischer U, Sarikaya H, Baumgartner RW, Georgiadis D, Odier C, Michel P, Putaala J, Griebe M, Wahlgren N, Ahmed N, van Geloven N, de Haan RJ, Nederkoorn PJ. Safety and functional outcome of thrombolysis in dissection-related ischemic stroke: a meta-analysis of individual patient data. Stroke 2011;42:2515-2520.
17. Villa A, Molgora M, Licari S, Omboni E. Acute ischemic stroke, aortic dissection, and thrombolytic

therapy. Am J Emerg Med 2003;21:159-160.
18. Sung PS, Fang CW, Chen CH. Acute aortic dissection mimicking basilar artery occlusion in a patient presenting with sudden coma. J Clin Neurosci 2010; 17:952-953.
19. Hiratzka LF, Bakris GL, Beckman JA, Bersin RM, Carr VF, Casey DE et al. 2010 ACCF/AHA/AATS/ ACR/ASA/SCA/SCAI/SIR/STS/SVM Guidelines for the Diagnosis and Management of Patients With Thoracic Aortic Disease. Circulation 2010;121:1544-1579.