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Case Report

Watershed cerebral infarction in a patient with a persistent primitive trigeminal artery and contralateral internal carotid artery stenosis: A case report☆,☆☆

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ABSTRACT

Patients with a persistent primitive trigeminal artery frequently have a poorly developed vertebrobasilar arterial system. However, they are not at higher risk of stroke and most are asymptomatic. Left cerebral watershed infarction was identified in a 75-year-old man who presented with aphasia and disorientation on magnetic resonance image (MRI). Additional imaging studies also demonstrated a right persistent primitive trigeminal artery, aplastic basilar artery, and 47% left internal carotid artery stenosis. Antiplatelet medication was administered and he was discharged 2 weeks after admission on aspirin. At the 4-month follow-up, cerebral blood flow in the left watershed territory was still decreased; however, no recurrent stroke had occurred. Although the indication for surgical or endovascular intervention for internal carotid artery stenosis is primarily determined by the degree of stenosis, cerebral blood flow evaluation is recommended in patients with internal carotid artery stenosis and a persistent primitive trigeminal artery.

Introduction

Persistent primitive trigeminal artery (PPTA) is an embryologic carotid–vertebrobasilar anastomosis identified on 0.1% to 0.6% of cerebral angiograms [1,2]. Most are unilateral and originate from the cavernous segment of the internal cerebral artery (ICA) near its posterior genu. The typical site of termination is the basilar artery (BA) between the superior cerebellar artery (SCA) and anterior inferior cerebellar artery (AICA) [3,4]. Hypoplasia of the proximal BA and lack of a posterior communicating artery (PComA) are frequently associated with PPTA [3]. PPTA does not cause symptoms in and of itself [5], and it does not increase the risk of cerebral in-

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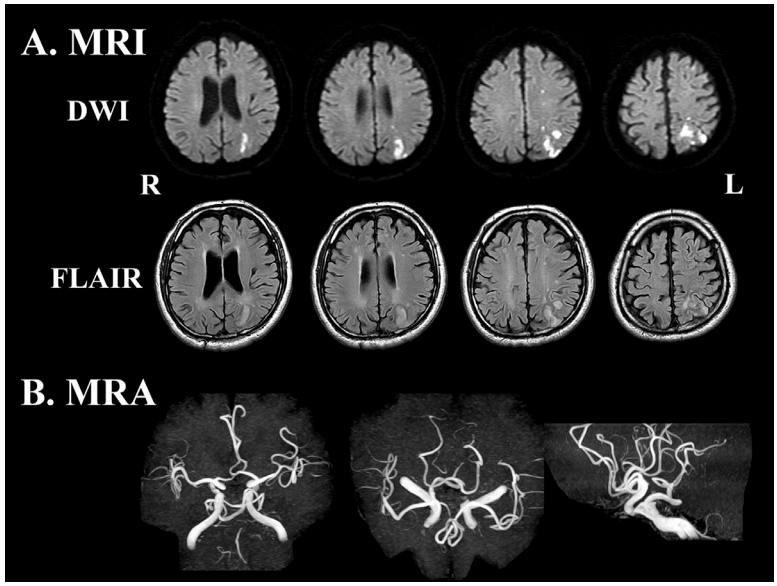

Fig. 1 – (A) Diffusion-weighted imaging (DWI) and fluid attenuated inversion recovery (FLAIR) imaging demonstrated high signal intensity lesions in the posterior cerebral–middle cerebral artery and anterior cerebral–middle cerebral artery watershed areas of the left parieto-occipital and frontal lobes. (B) No intracranial artery stenosis was recognized on magnetic resonance angiography (MRA). However, a large persistent primitive trigeminal artery originated from the cavernous portion of the right internal carotid artery and terminated on the distal portion of the basilar artery, presumably supplying blood to the posterior circulation. The basilar artery was aplastic proximal to its junction with the primitive artery and both vertebral arteries were hypoplastic. Posterior communicating arteries were not identified on either side.

farction. In patients with a PPTA, the brainstem is the most common site of infarction, even when the cause is ICA stenosis or dissection. This is because an ICA embolus can migrate to the posterior circulation via the PPTA [6,7]. Hemodynamic-induced cerebral infarction is rare in patients with a PPTA, and to the best of our knowledge, only a single case of watershed infarction associated with PPTA has been previously reported [8]. We present a patient with ICA stenosis and a PPTA who experienced a watershed infarction owing to decreased cerebral blood flow (CBF) which was demonstrated on perfusion computed tomography (CT). This case demonstrates the importance of CBF evaluation in patients with ICA stenosis who also have a PPTA.

Case presentation

A 75-year-old man experienced the sudden inability to type words while working on a computer. The next day, his wife took him to our hospital. On examination, he was disoriented with right–left confusion, finger agnosia, acalculia, and agraphia. Motor testing was normal. Blood pressure was 167/96 and heart rate was 88 beats/minute without arrhythmia. Diffusion weighted imaging (DWI) of magnetic resonance imaging (MRI) showed high signal intensity lesions in the watershed areas of the left parieto-occipital and frontal lobes. These lesions were also identified on fluid attenuated inversion recovery (FLAIR) imaging (Fig. 1A). Magnetic resonance angiography (MRA) demonstrated a Saltzman type I PPTA which arose from the cavernous portion of the right ICA and connected to the distal BA [9], aplasia of the BA proximal to its junction with the PPTA, and hypoplasia of the vertebral arteries (VAs). The anterior communicating artery was intact but the A1 segment of the right anterior cerebral artery (ACA) was narrow. Neither a right nor left PComA was identified on MRA (Fig. 1B). The patient was diagnosed with type 2 diabetes mellitus and hyperlipidemia but had discontinued taking the corresponding medications on his own initiative 1 year previously. Laboratory testing was as follows: serum glucose, 223 mg/dL; HbA1c, 11.0%; total cholesterol, 227 mg/dL; low-density lipoprotein cholesterol, 124 mg/dL; high-density lipoprotein cholesterol, 37 mg/dL; and triglyceride, 329 mg/dL. Twelve-lead electrocardiography and continuous electrocardiographic monitoring did not show atrial fibrillation at any time during his admission. Treatment with an intravenous argatroban hydrate infusion and oral aspirin was initiated. Three-dimensional CT angiography demonstrated aplasia of the BA proximal to the PPTA junction, hypoplasia of the right A1 segment of the right ACA, the PPTA branching off the ICA, and 47% left ICA stenosis (North American Symptomatic Carotid Endarterectomy Trial criteria); carotid ultrasonography also showed moderate (49.7%) stenosis of the left ICA (Fig. 2).

The patient's right–left confusion and finger agnosia improved in 3 days, and acalculia and agraphia in 10 days. A right inferior quadrantanopia was identified on visual field testing. Two weeks after admission, he was discharged on aspirin and rosuvastatin. At the 4-month follow-up, his laboratory data had improved (serum glucose, 102 mg/dL; HbA1c,

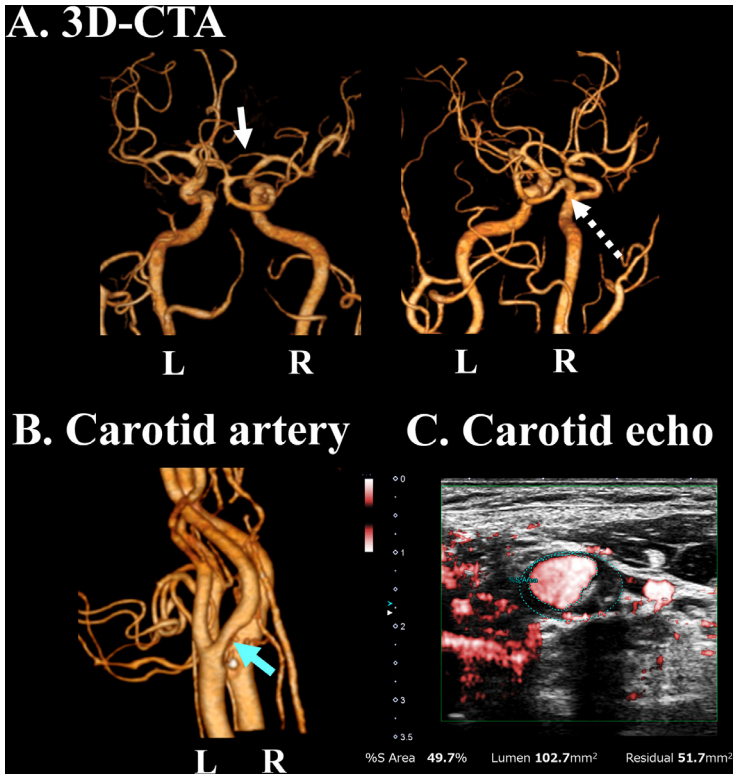

Fig. 2 – (A) On three-dimensional computed tomography angiography, the A1 segment of the right anterior cerebral artery was hypoplastic (white arrow). The left vertebral artery was identified but the basilar artery was aplastic. A persistent primitive trigeminal artery originating from the cavernous portion of the right internal carotid artery supplied blood flow to the distal basilar artery (dotted white arrow). (B) Moderate stenosis was identified in the left cervical internal carotid artery just distal to the bifurcation (blue arrow). (C) Carotid ultrasonography showed 49.7% stenosis of the left cervical internal carotid artery.

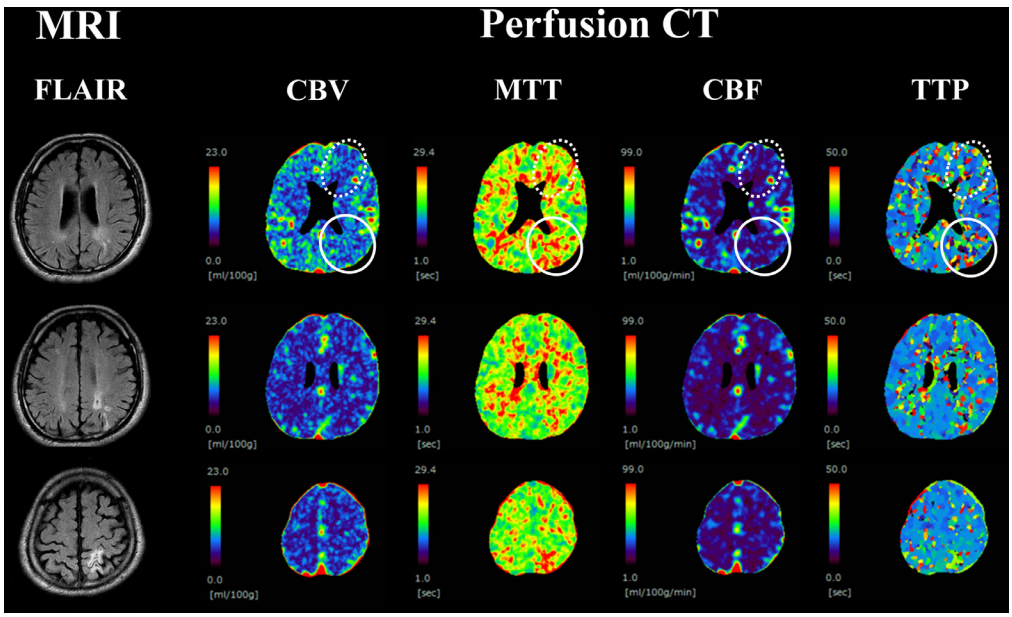

Fig. 3 – At the 4-month follow-up, perfusion computed tomography demonstrated decreased cerebral blood volume (CBV) and flow (CBF) and delayed mean transit time (MTT) and time-to-peak (TTP) over a wider area than the areas of infarction on FLAIR imaging. The dotted white circles indicate the watershed area between the anterior cerebral and middle cerebral arteries; the solid white circles indicate the watershed area between the middle cerebral and posterior cerebral arteries.

7.1%; total cholesterol, 173 mg/dL, low-density lipoprotein cholesterol, 101 mg/dL; high-density lipoprotein cholesterol, 46 mg/dL; and triglyceride, 131 mg/dL). FLAIR imaging demonstrated high signal intensity areas consistent with completed infarction, and perfusion CT showed decreased cerebral blood volume (CBV) and CBF and delay of mean transit time (MTT) and time-to-peak (TTP) in the left watershed areas that is larger than the completed infarction areas (Fig. 3). The patient elected to continue medical therapy rather than proceed with surgical or endovascular treatment of the ICA stenosis.

Discussion

PPTA is usually an incidental finding on MRA because the anomalous artery does not cause symptoms in and of itself [5]. PPTA is typically associated with a poorly developed vertebrobasilar arterial system [3,9]. In one PPTA study, hypoplasia of the vertebrobasilar artery system was severe in 28% of cases, moderate in 47%, and absent in 26% [1]; the authors speculated that the more the PPTA serves as the main blood supply to the posterior circulation, the more hypoplastic the VA and proximal BA become. In our patient, the PPTA seemed large enough to supply sufficient blood flow to the SCA and the PCA. The vertebrobasilar system was not supplying these arteries because the proximal BA was aplastic and the VAs were hypoplastic. Therefore, the entire blood supply to the brain was provided only by the ICAs. Although no stenosis was seen in the MCAs, ACAs, or PCAs, the blood supply to the distal cortex on the left appeared insufficient on perfusion CT owing to the left ICA stenosis. Right A1 hypoplasia and absence of both PComAs also contributed to poor left watershed territory blood supply.

The main indication for surgical or endovascular treatment of ICA stenosis is based on the degree of stenosis, and CBF is not always evaluated. In asymptomatic patients, internal carotid endarterectomy or stenting is indicated in those with ≥70% stenosis [10]. Although our patient had only a 47% ICA stenosis on computed tomography angiography, watershed infarction occurred because of low blood flow. After 4 months of antiplatelet therapy, stroke has not recurred. However, antiplatelet medication does not address CBF, and the reduced blood flow in the watershed areas on perfusion CT suggests he is still at considerable risk for another watershed infarction. Therefore, in patients with a PPTA and severely hypoplastic vertebrobasilar system, the authors recommend a CBF imaging study to determine if surgery is indicated, as well as imaging to evaluate the degree of ICA stenosis.

Patient consent

The patient provided written consent for publication of this case report and any accompanying images.

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