A 46-year-old woman without cardiovascular risk factors who was taking oral prednisolone for Sjögren syndrome presented with repeated episodes of transient left hemiparesis and left-sided sensory disturbance over a 2-year period despite preventive therapy with antiplatelets and anticoagulants. Infarcts were always identified in the right middle cerebral artery territory on brain magnetic resonance imaging (Figure 1) without any sign of cerebrovascular stenosis or embolicogenic sources. She was referred and admitted to our hospital 2 years after the initial onset. Carotid magnetic resonance angiography showed that the origin of the right internal carotid artery (ICA) bent inward (Figure 2A). However, follow-up magnetic resonance angiography the next day revealed that the origin of the right ICA bent outward, as commonly seen (Figure 2B). On carotid ultrasonography, the origin of the right ICA was seen to move dynamically according to head rotation and swallowing (Movie I in the online-only Data Supplement). The origin of the right ICA shifted to the inner side of the right external carotid artery and became stuck with head rotation to the left and swallowing. Conversely, the position of the origin of the right ICA returned to the outer side of the right external carotid artery with head rotation to the right and swallowing. In addition, a mural thrombus was detected at the origin of the right ICA on carotid ultrasonography (Figure 3). The thrombus gradually reduced and disappeared with continuous intravenous infusion of argatroban for 4 days. Cervical T1-weighted magnetic resonance imaging revealed that the origin of the right ICA was hooked by the right greater cornu of the hyoid bone when it became stuck (Figure 4). Dynamic movement of the origin of the right ICA with head rotation and swallowing in daily life and frequent mechanical stimulation by the hyoid bone appeared to result in endothelial injury, thrombus formation, and artery-to-artery embolism to the brain. Long-term steroid therapy might have enhanced the prothrombic state and could thus have contributed to thrombus formation.

Partial resection of the right greater cornu of the hyoid bone and carotid endarterectomy were performed. Intraoperatively, the vessel wall of the origin of the right ICA was thickened with ulceration (Figure 5). Microscopically, the specimen obtained by carotid endarterectomy demonstrated medial necrosis with inflammatory changes and mural organized thrombus without atherosclerosis. The postoperative course was uneventful. She was discharged with no neurological deficit and remained free of ischemic attack at >6 months postoperatively.

Compression of the carotid artery by the hyoid bone was reported as a cause of carotid steno-occlusive disease.1–4 A unique finding in the present case compared with previous reports is that dynamic movement of the origin of the right ICA enhanced injury by the hyoid bone that was causing artery-to-artery embolism. The life-threatening embolicogenic lesion was accurately detected by complex vascular imaging studies and successfully removed by surgical treatment.

Disclosures

None.

References

Figure 1. Brain diffusion-weighted imaging demonstrating fresh infarcts in the right middle cerebral artery territory (arrows).

Figure 2. Carotid magnetic resonance angiography demonstrating dynamic movement of the origin of the right internal carotid artery (ICA). A, The origin of the right ICA is seen bending inward (arrows). B, The next day, the origin of the right ICA is seen bending outward (arrowheads).

Figure 3. Longitudinal B-mode imaging on carotid ultrasonography demonstrating a mural thrombus (arrow) at the origin of the right internal carotid artery.

Figure 4. Cervical T1-weighted imaging demonstrating the origin of the right internal carotid artery (arrow) hooked by the right greater cornu of the hyoid bone (arrowheads).
Figure 5. Intraoperative view demonstrating the vessel wall of the origin of the right internal carotid artery thickened with ulceration (arrow). CCA indicates common carotid artery; ECA, external carotid artery; and ICA, internal carotid artery.
Repellite Artery-to-Artery Emboli Formation by Dynamic Movement of the Internal Carotid Artery and Mechanical Stimulation by the Hyoid Bone
Keisuke Tokunaga, Toshiyuki Uehara, Hideki Kanamaru, Hiroharu Kataoka, Kozue Saito, Hatsue Ishibashi-Ueda, Ryogo Shobatake, Yoshifumi Yamamoto and Kazunori Toyoda

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