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SUMMARY An unusual case of peripheral hypoglossal nerve palsy, caused by lateral position of the external carotid artery and an abnormally high carotid bifurcation is reported. Improvement followed ligation and cutting of the external carotid artery at its origin.

VARIATIONS AND ANOMALIES are common at the bifurcation of the external and internal carotid arteries.1 Lateral position of the external carotid artery2–5 formerly considered extremely rare, has recently been found to be more common.5,6 However, there are few reports, that this condition causes neurological signs.1 In this paper, a case of peripheral hypoglossal nerve palsy caused by lateral position of the external carotid artery and a carotid high bifurcation is presented.

Case Report

A 49-year-old man was admitted because of tongue deviation and dysarthria on December 8, 1980. At age 42, his blood pressure was found to be high and anti-hypertensive drugs were presented. He had felt throbbing pains in the occipital region beginning November 10, 1980, but these pains became intense from midday of November 21. At that time he visited a neurosurgical clinic, but no abnormalities could be found. On the morning of November 22, eating became difficult, and movement of the tongue was impeded, making speech inarticulate.

Tongue deviation to the right and atrophy and fasciculation of the right lingual muscle were observed (fig. 1). Tactile sensibility and taste sensation of the tongue were normal, and no abnormality of other cranial nerves was observed. There were no other abnormalities on neurological examination. Horner’s sign was not presented and no bruit was heard in the neck. A diagnosis of peripheral hypoglossal nerve palsy with suspected hypoglossal neurinoma was made.

A CT scan demonstrated no abnormality related to the right hypoglossal palsy. Right jugular venography revealed good filling by contrast medium and no abnormal findings. Tomography of the hypoglossal canals, was normal. Bilateral carotid angiography and bilateral vertebral angiography were performed. No abnormalities were observed in the vertebral angiogram, except conspicuous elongation of the basilar artery. In the right carotid angiogram, marked lateral displacement of the right external carotid artery was noticed and the bifurcation of the external and internal carotid arteries was abnormally high (upper margin of the C1 vertebra); there was conspicuous kinking and elongation of the external and internal carotid arteries and, in particular, prominent dilatation of the proximal portions of both arteries. In the lateral view of the angiogram, a loop-like elongation was observed 3.5 cm distal to the bifurcation (fig. 2). In the left carotid angiogram, as in that of right side, the bifurcation was at a high level (center of C2 vertebra) and elongation was conspicuous, but a lateral position of the external carotid artery was not seen.

From this examination, the possibility of hypoglossal neurinoma was excluded and the following two mechanical processes were considered as cause of right peripheral hypoglossal nerve palsy:

1) Pressure on the hypoglossal nerve owing to the lateral position of the external carotid artery, and an abnormally high position of the bifurcation of the internal and external carotid arteries.

2) Pressure owing to loop-like elongation of the internal carotid artery 3.5 cm distal to its bifurcation.

It was considered, however, that if the hypoglossal nerve palsy was caused by change around the loop-like elongation of the distal internal carotid artery, it might well be accompanied by a paralysis of the glossopharyngeal, vagus or accessory nerve running closer to the internal carotid artery than the hypoglossal nerve. Therefore, the hypoglossal palsy more likely was due to the lateral position of the external carotid artery and the abnormally high position of the bifurcation.

Surgery of the right of the neck was performed on March 23.

Operative findings: Figure 3 shows the photograph of the neck taken during the operation. As seen, the hypoglossal nerve was running just above the bifurcation of the internal and external carotid arteries. The hypoglossal nerve was markedly compressed and extended laterally by the lateral position of the external carotid artery and the tortuous dilatation of the proximal portions of the external and internal carotid arter-
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Figure 1a. Preoperative picture of the patient. The tongue showed right atrophy and fasciculation.

ies. Although the sternomastoid branch of the occipital artery slightly compressed the hypoglossal nerve, this compression was not considered to be the cause of nerve palsy. Around the bifurcation, there were several lymphnodes of about 1-2 cm diameter which somewhat increased the tension on the hypoglossal nerve. These lymphnodes were removed and the external carotid artery was tied and cut at its origin. The sternomastoid branch was cut from the occipital artery. The superior thyroid artery was preserved.

Postoperatively, the hypoglossal nerve was able to cross the internal carotid artery without strain and with some bend. The lymphnodes, which was removed during operation, showed non-specific inflammation.

Postoperatively the right deviation of the tongue began to improve from the second day after the operation, and a week late, dysarthria was hardly noticeable. The patient was discharged on April 25. 22 months after the operation, atrophy of the right lingual muscle is hardly noticeable (fig. 1b).

Discussion

There are several recent reports of cases of peripheral hypoglossal nerve palsy due to vascular en-

Figure 2. Right carotid angiogram. Lateral position of the external carotid artery is seen in the A-P view. Dilatation and a tortuosity of the proximal portion of the external and internal carotid arteries are also seen. The bifurcation of the external and internal carotid arteries are at the upper margin of the C2 vertebra. In the lateral view, loop formation of the dilated internal carotid artery is seen 3.5 cm distal to its origin. CC = common carotid artery; EC = external carotid artery; IC = internal carotid artery; VA = vertebral artery; ST = superior thyroid artery; L & F = Lingual & facial arteries; O = occipital artery; AP = posterior auricular artery; TS = superficial temporal artery; M = maxillary artery.
FIGURE 3. Appearance at operation. The hypoglossal nerve crosses the proximal portion of the external and internal carotid arteries. The dilated and tortuous bifurcation compresses the XIIth nerve laterally. (1) = external carotid artery; (2) = hypoglossal nerve; (3) = common carotid artery; (4) = sternomastoid branch of the occipital artery; (5) = superior thyroid artery; (6) = digastric muscle; Lymphnodes around the bifurcation have already been removed in this picture.

Entrapment caused by kinking and elongation of the internal carotid artery and the sternomastoid artery. In these cases, the hypoglossal nerve was compressed by an abnormal loop of the internal carotid artery and was fixed and grooved by the sternomastoid branch of the occipital artery. It is also known that hypoglossal nerve palsy sometimes occurs after carotid endarterectomy, and entrapment of the sternomastoid artery was suspected in these cases. There is also a report of peripheral hypoglossal nerve palsy due to a dural AVM which caused formation of abnormal vascular nets in the hypoglossal canal. Thus structures in the neck, especially the internal carotid artery, external carotid artery and their branches may cause peripheral hypoglossal nerve palsy. But, as mentioned above, there are few reports of peripheral hypoglossal nerve palsy attributable to an abnormal vascular condition.

The first report of a lateral position of the external carotid artery was that of Hyrtl in 1841. Since then, Yuhina, Lie, Schreiber, Handa and Ojemann have reported this phenomenon. Seidel and Lie showed this abnormality by angiography in 1965, and 1968. Statistical investigations of the lateral position were made by Teal in the USA and Tatezawa in Japan. We studied the incidence of lateral position of the external carotid artery in 672 consecutive carotid angiograms over the past two years findings that the incidence was 4.9% (7.9% on right side, 2.3% on left side and 1.0% on both sides) which was similar to the value reported by Tatezawa, but not so high as that reported by Teal. Lateral position of the external carotid artery is not uncommon. The right side was 3.4 times more affected than the left.

The height of the bifurcation of the external and internal carotid arteries is shown in figure 4. These data were obtained from 450 carotid angiograms (50 cases each in decades from 0 to 8) performed in 1980 in our hospitals. In the 3rd decade, the average position of the bifurcation was the center of vertebra C3. In the present case, the position of bifurcation was at the upper margin of the C2 vertebra, which was markedly higher than the average.

What was the cause of the palsy in this case? Usual-
The hypoglossal nerve runs out from the hypoglossal canal, and passes down to the posterior part of the internal carotid artery, crossing it 1–2 cm above the bifurcation of the internal carotid and external carotid arteries. In this case, owing to the lateral position of the external carotid artery and the abnormally high position of its bifurcation, the hypoglossal nerve was forced to make a long detour on the outside of the external and internal carotid arteries. Moreover, it was compressed toward the outside so much that there was conspicuous kinking, elongation and especially tortuous dilation in the proximal portions of these arteries. The bifurcation might have been pushed upward owing to the loop-like elongation of the internal carotid 3.5 cm distal to the bifurcation and the hypoglossal nerve might have been forced to run along the thickest part of the bifurcation of the internal and external carotid arteries.

It seems probable that a vascular compression of the hypoglossal nerve and paralysis were caused by the hemodynamic pressure and the vibration arising from arterial pulsation. Swelling of the lymphnodes, which was observed at the time of the operation, might also have contributed to the paralysis.

When, as in the case, the hypoglossal nerve palsy is attributable to vascular compression by the arteries in the neck, it can be cured by a relatively simple operation, and the prognosis is good in most cases. Therefore, in cases of peripheral hypoglossal nerve palsy, in which the responsible focus cannot be found by examinations such as CT scans, further examination should be performed by carotid angiography on the paralyzed side.
Peripheral hypoglossal nerve palsy caused by lateral position of the external carotid artery and an abnormally high position of bifurcation of the external and internal carotid arteries--a case report.
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