

## 이분 정중신경을 동반한 잔류 정중동맥 폐색에 의한 급성 수근관 증후군: 증례 보고

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## Thrombosed Persistent Median Artery with Bifid Median Nerve Causing Acute Carpal Tunnel Syndrome: A Case Report

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Among the numerous causes of carpal tunnel syndrome, thrombosed persistent median artery with bifid median nerve is extremely rare. Our presentation shows that an unusual case and surgical outcome of thrombosed persistent median artery with bifid median nerve causing acute carpal tunnel syndrome. Care has to be taken that identification of abnormal anatomy in the carpal tunnel is essential, especially in abruptly occurred carpal tunnel syndrome. We suggest that Doppler ultrasound examination is very useful in an outpatient clinic-based for identification of abnormal structure, which makes more quickly and accurate diagnosis than magnetic resonance image.

**Key Words:** Carpal tunnel syndrome, Persistent median artery, Bifid median nerve, Occlusion

Carpal tunnel syndrome is the most common compressive neuropathy of the upper extremity<sup>1</sup>. Moreover, several causative factors increase the carpal tunnel pressure, which include space-occupying mass, tenosynovitis, aberrant musculature, or congenital abnormal median nerve. A persistent median artery is a numerous reported anatomical variant, which is supposed to degenerate over time<sup>2,3</sup>. In addition, studies have shown its co-existence with bifid median nerve. Most cases of persistent median artery are known to be asymptomatic<sup>4</sup>, although some re-

ports demonstrated that co-existence of persistent median artery with bifid median nerve can lead to carpal tunnel syndrome<sup>5</sup>. However, a case of thrombosed persistent median artery with bifid median nerve is extremely rare. Hence, we report an unusual case and surgical outcome of thrombosed persistent median artery with bifid median nerve causing acute carpal tunnel syndrome. Informed consent was obtained from the patient for purpose of the case report.

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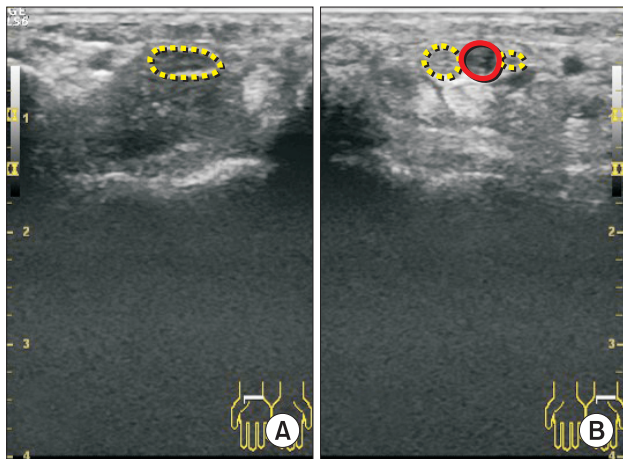
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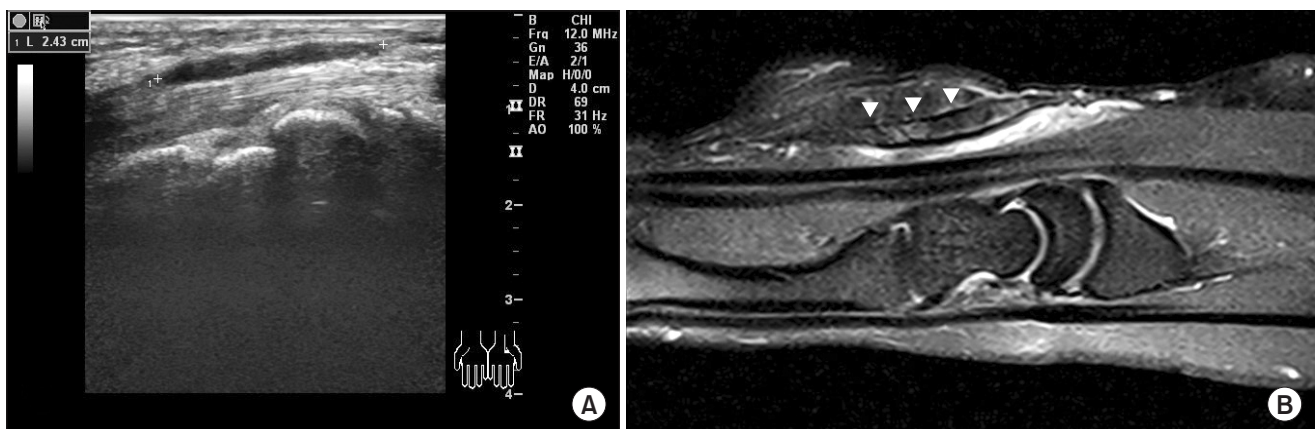
## CASE REPORT

A 58-year-old female patient was referred to corresponding author's hospital for pain at the volar aspect of the left wrist and paresthesia with sensory loss at the median nerve territory from the thumb to the middle finger. The patient had no prior history of taking anti-diabetic, anti-hypertensive, and any thrombogenic-related medications. The chief complaint was pain that occurred acutely, and the symptoms persisted for five weeks. The patient's numbness was not relieved by oral medication. On physical examination, both Tinel sign and Phalen test were

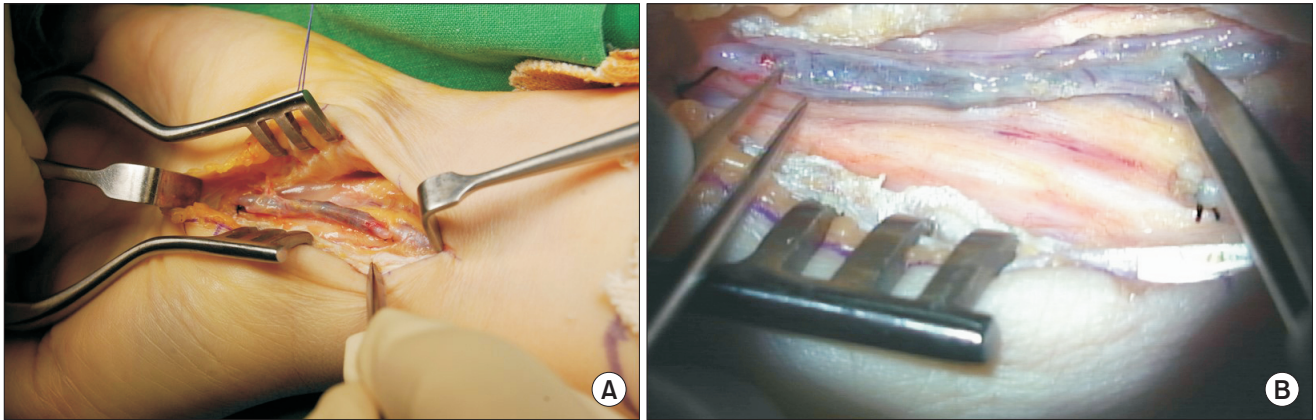


**Fig. 1.** The Doppler ultrasound. (A) Normal echogenicity and contour of the median nerve (dotted circle) in the healthy wrist. (B) Persistent median artery (lined circle) between the divided median nerves (dotted circles) in the affected wrist.

positive. However, no thenar muscle atrophy was noted. Although the electrodiagnostic study at corresponding author's hospital revealed no specific abnormality, we were able to identify a persistent median artery between the divided median nerves on Doppler ultrasound examination (Fig. 1). The persistent median artery was completely obstructed in the carpal tunnel area, but blood flow was identified to be proximal to the carpal tunnel. In addition, we identified a thrombus — that was approximately 4 cm in length — of the persistent median artery at the wrist on both the magnetic resonance image and Doppler ultrasound (Fig. 2). The surgery was performed with an operating microscope. First, we performed open carpal tunnel release. The skin was incised using a curved longitudinal incision, passing the wrist crease. Subsequently, we could identify the split median nerve from the inlet of the carpal tunnel, which is proximally 2 cm. In addition, the radial-side branch of the median nerve was more dominant than the other side, and the persistent median artery was found between the split median nerve. The persistent median artery was completely obliterated in 5 mm diameter, and a 2-cm length of the artery was obliterated proximally and distally from the carpal tunnel inlet. Under the operating microscope, the obstructed artery was resected and ligated proximally (Fig. 3). The patient had previous paresthesia of the affected hand, which was resolved immediately postoperatively, and preoperative chief complaint was completely resolved at 9 months postoperatively.



**Fig. 2.** (A) The thrombosis in about 4 cm length of the persistent median at the wrist on the ultrasound examination. (B) The thrombosis was indicated with arrowhead (arrowheads) on magnetic resonance image.



**Fig. 3.** (A) The persistent median artery was completely obliterated in each 2 cm length proximally and distally from the carpal tunnel inlet. (B) Under the operating microscope, the obstructed artery was resected and ligated proximally.

## DISCUSSION

Carpal tunnel syndrome has typical symptoms, including paresthesia or numbness in the region of median nerve distribution<sup>1</sup>. However, patients typically do not complain about symptoms on the volar aspect of the wrist. If the patient has these symptoms, we should rule out other uncommon causes of compressive neuropathy of the wrist. Acute onset of median nerve entrapment symptom tends to result from trauma, swelling, infections, inflammation, anomalous anatomy, coagulopathy, or tumorous conditions<sup>6</sup>. In this present case, atypical symptoms were noted, which were painful sensation at the volar aspect of the wrist and abrupt symptom onset. These are the reasons why we performed further evaluation for the possibility of abnormal anatomy or mass-like lesion.

The incidence of bifid median nerve varies from 0.8% to 21.0% in patients with carpal tunnel syndrome<sup>7</sup>. The various differences of the incidence maybe because of various diagnostic tools or ethnic groups. In addition, several reports have suggested that the bifid median nerve can be a causative factor for carpal tunnel syndrome<sup>8</sup>. One of the hypotheses is that the bifid median nerve may facilitate compression of the nerve in the carpal tunnel because of larger summated cross-sectional area compared with the cross-sectional area of a non-bifid median nerve<sup>8</sup>. In contrast, some authors recently reported that the bifid median nerve is not an independent risk factor

for development of carpal tunnel syndrome<sup>7,9</sup>.

Meanwhile, co-existence of the bifid median nerve and persistent median artery was suggested by several authors<sup>2,9</sup>. Embryologically, the upper limb develops at the end of four weeks to eight weeks gestation. Angiogenesis of the upper limb is followed with development of the upper limb. Briefly, the median artery is the main branch of the brachial artery at the forearm area at first, followed by the ulnar and radial arteries that play a major role for providing the blood supply below the forearm level while the median artery degenerates<sup>2</sup>. At this time, the persistent median artery can cause splitting of the median nerve, which is thus called bifid median nerve. Therefore, when we encounter either the bifid median nerve or persistent median artery, we should evaluate for the co-occurrence of both anatomical abnormalities. Generally, two imaging modalities can identify an abnormal finding in the carpal tunnel, the Doppler ultrasound and magnetic resonance imaging. Most of the cited reports have used a Doppler ultrasound for diagnosis of persistent median artery and bifid median nerve in the carpal tunnel. Based on our present case, we also recommend that the Doppler ultrasound examination is useful for identification of this abnormal anatomy in the carpal tunnel area in the outpatient clinic. In addition, a clear understanding of the abnormal anatomy is very important when performing surgery for carpal tunnel syndrome.

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syndrome can be conservative or surgical treatment. Kele et al.<sup>5</sup> reported that surgical management of this case produced a good outcome. In contrast, Salter et al.<sup>10</sup> considered treatment with anticoagulants as a conservative management. Although he did not mention the accurate period of symptom duration, the report suggested early detection of the thrombosed median artery can be of great help for preventing the need for any surgery or thrombolytics. Our present case also demonstrated an acute onset of compressive median nerve neuropathy, but the symptoms of obliterated median artery had persisted for five weeks. That is why we considered surgery for the management of the obstructed median artery and compressive median nerve neuropathy. Based on our experiences, we think that a problematic vessel in the past can also tend to be problematic in the future, and the carpal tunnel space and proximal area are sites vulnerable to compression of the contents in the carpal tunnel space anatomically. The persistent median artery can also be easily compressed around the carpal tunnel. Hence, we recommend surgical management for this abnormal finding rather than a conservative treatment in the same case, although immediate medical therapy can help avoid surgery in the early stage.

In conclusion, careful identification of an abnormal anatomy in the carpal tunnel is essential, particularly in cases wherein carpal tunnel syndrome occurred abruptly. Among the diagnostic tools, Doppler ultrasound examination is very useful in an outpatient clinic, because it provides a quicker and more accurate diagnosis than magnetic resonance imaging. In addition, we suggest that surgical treatment for a thrombosed persistent median artery in carpal tunnel syndrome provides a good result.

## CONFLICTS OF INTEREST

The authors have nothing to disclose.

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## 이분 정중 신경을 동반한 잔류 정중동맥 폐색에 의한 급성 수근관 증후군: 증례 보고

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수근관 증후군(carpal tunnel syndrome) 원인 중 드물게 잔류 정중동맥(persistent median artery)에 의한 수근관내 정중 신경의 압박이 보고되어 있다. 잔류 정중동맥은 비교적 많이 보고된 해부학적 변이이지만, 이분 정중 신경을 동반하며 혈전으로 인한 잔류 정중동맥의 폐색이 급성 수근관 증후군을 초래한 증례는 굉장히 드문 것으로 보고된다. 본 증례는 이분 정중 신경을 동반한 잔류정중동맥의 폐색에 의한 급성 수근관 증후군 사례로 수근관 증후군을 수술하는 외과의라면 손목통증을 동반한 신경증상이 있는 경우 술 전 초음파 및 도플러검사의 중요성을 염두에 두어야 할 것이다.

**색인단어:** 수근관 증후군, 잔류 정중동맥, 이분 정중 신경, 폐색

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