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A Rare Case of Guyon's Canal Syndrome Caused by Cystic Adventitia Degeneration: High-Resolution Ultrasound Findings

Introduction

Entrapment of the ulnar nerve inside the ulnar tunnel, which is also called Guyon's canal, is a well-known entity leading to pain, numbness and loss of function in the part of the hand supplied by the ulnar nerve. Space-occupying lesions in this area, such as ganglions, tendinitis, fracture, arthritis, nonspecific edema and thromboangitis, may cause the entrapment of the nerve, known as ulnar tunnel syndrome.

Cystic adventitial disease (CAD) is a rare benign vascular lesion, where cysts form in the adventitia of a blood vessel, first described in the iliac artery (Atkins HJ, Key JA. *Br J Surg* 1947; 34: 426). CAD may cause local pain due to the stenosis and occlusion of the vessel and can be treated either with US- or CT-guided percutaneous cyst aspiration or surgery (Drac P et al. *Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub* 2011; 155: 309–321).

For unknown reasons, most of the CAD cases reported thus far involve the popliteal artery in young middle-aged men (Drac P et al. *Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub* 2011; 155:309–321). A few cases have also been described in the external iliac, common femoral, axillary, distal brachial, and radial arteries.

Although the pathomechanisms of CAD remain controversial, a theory of joint-related origin has been suggested, similar to the pathogenesis of intraneural ganglion cysts (Spinner RJ et al. *Clin Anat* 2013; 26: 267–281). The diagnosis can be established with high-resolution ultrasound and MRA as the first-choice imaging methods (Drac P et al. *Biomed Pap Med Fac Univ Palacky Olomouc Czech Repub* 2011; 155: 309–321).

We report here a unique case of cystic adventitial degeneration that affected the ulnar artery and caused ulnar nerve palsy, which was detected with high-resolution ultrasound.

Case Presentation

A 52-year-old man complained of paresthesia in the area of the fourth and fifth finger of his left hand for the last two months. On clinical investigation, he showed restricted mobility and atrophy of the adductor pollicis and lumbrical muscles between the fourth and fifth fingers. He also had a positive "Tinel's sign" at the Guyon's canal.

He was referred to the radiology department for an ultrasound examination of the right wrist. Ultrasound was performed with a broadband linear array using 18 MHz (Logic e9, GE General Electric, Milwaukee, USA).

In the "loge de Guyon" we found compression of the ulnar nerve due to a cystic lesion arising from the wall of the ulnar artery. The cystic lesion demonstrated a semicircular shape within the arterial wall (► **Fig. 1**) and more distally showed a connection to the articular capsule of the wrist joint (► **Fig. 1 d**). The subsequent MRI suggested vasculitis. Digital subtraction angiography did not reveal any abnormalities.

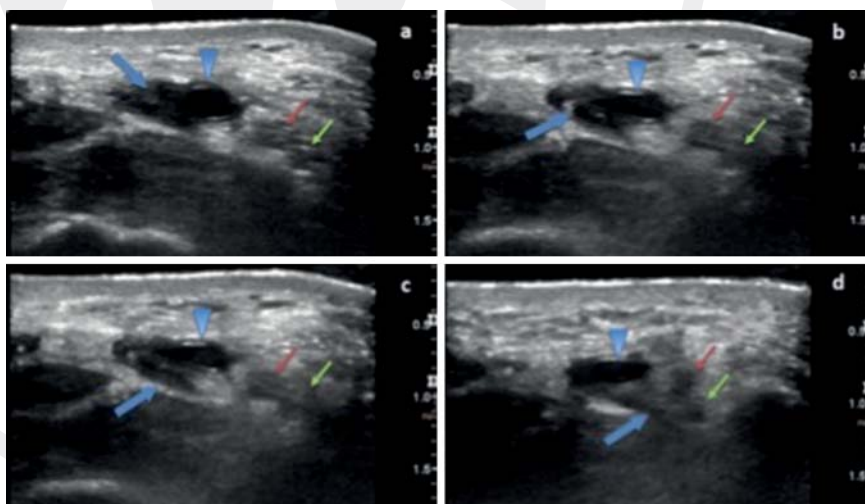
The patient then underwent surgery and the surgical findings confirmed a cystic

lesion within the ulnar artery wall that had a connection to the carpal joint (► **Fig. 2**). The swollen ulnar nerve was released and the cystic lesion was removed without resecting the whole ulnar artery. Histological findings revealed that the lesion consisted of mucoid tissue.

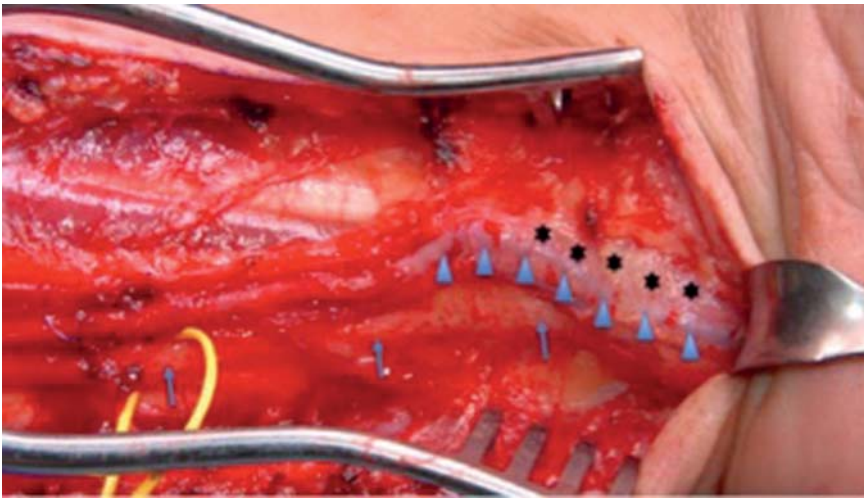
The patient recovered from the ulnar palsy within a week of the operation. Follow-up after several weeks did not reveal any recurrence of symptoms.

Discussion

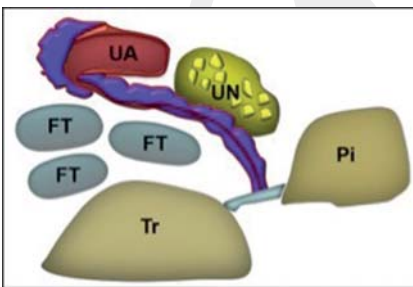
In this study, we report a patient who came to our hospital emergency department with ulnar tunnel syndrome that was caused by a cystic mass inside the ulnar artery wall. To our knowledge, this is the first time CAD has been described in the ulnar artery, as well as the fact that it can lead to direct compression of the ulnar nerve with associated consequences. It is known that regular ganglions emerging from the wrist joint may compromise the ulnar nerve in Guyon's canal (Zielinski CJ. *Orthopedics* 2003; 26: 429–430). In our case, however, the cystic lesion may have started to form inside the blood vessel



► **Fig. 1** Consecutive transverse ultrasound scans in the wrist area from proximal to distal **a-d** show a cystic mass (fat arrows) that partly surrounds the radial artery (arrow heads) and is indistinct from the artery wall **a, b**. More distally, this mass has a connection (fat arrow) to the wrist joint **c, d** and is in contact with the superficial and deep branches of the ulnar nerve (red and green arrows, respectively).



► **Fig. 2** Photograph taken during the operation shows the ulnar artery (arrow heads) surrounded by cystic tissue (asterisks) next to the ulnar nerve (arrows).



► **Fig. 3** Schematic figure showing the cystic mass inside the ulnar artery adventitia that continues along a wall of the small articular branch to the wrist joint. (UA = ulnar artery, UN = ulnar nerve, FT = flexor tendon, Pi = pisiform, Tr = triquetrum).

adventitia of a small articular branch and progressed along the branch more proximally into the wall of the ulnar artery (► **Fig. 3**). Indeed, the connection to the wrist joint seen on ultrasound seems to support the theory of an articular origin of such lesions (Spinner RJ et al. *Clin Anat* 2013; 26: 267 – 281).

A recent report described a CAD in the radial artery with neurological symptoms of the median nerve. However, these were probably more incidental as the adventitial cysts were not in direct contact with the nerve (Wang HR et al. *J Hand Surg Am* 2007; 32: 126 – 130).

Due to the fact that the ulnar nerve and the ulnar artery run very close together in the narrow Guyon's canal, it is even more important to consider this pathology when confronted with an ulnar tunnel syndrome of unknown origin. Therefore, we suggest that cystic adventitial degeneration of the ulnar artery wall should be added to the more well-known causes of ulnar tunnel syndrome, e. g., tendinitis, ganglions, fracture, arthritis, and thromboangitis. As seen in our case, when carried out by an experienced radiologist, CAD can be diagnosed by high-resolution US as the first imaging study, and should reveal all the details necessary for the surgeon. Additional MRI or angiography may help further to identify the origin of the

ganglion, show the disturbance of the artery and the nerve, and clarify the anatomical situation for the surgeon, although, in our case, neither of these modalities provided additional information and angiography failed to show the abnormality. In conclusion, we can report that, even if CAD in the ulnar artery is probably a very rare cause for an ulnar tunnel syndrome, it may be under-recognized and, therefore, should be considered when investigating the wrist.

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