folia medica

Folia Medica 64(3):532-536
DOI: 10.3897/folmed.64.e62461

Case Report

Clinical-Electrodiagnostic Correlates in Three Cases of Riche-Cannieu Anomaly (All-Ulnar Hand)

Abdullah Alajmi[1], Adnan Khuraibet[1], Todor Shamov[2], Rossen Rousseff[1,3]

[1] *Department of Neurology, Ibn Sina Hospital, Kuwait city, Kuwait*

[2] *Department of Neurosurgery, Military Medical Academy, Sofia, Bulgaria*

[3] *Department of Neurology and Neurosurgery, Medical University of Pleven, Pleven, Bulgaria*

Corresponding author: Rossen T. Rousseff, Department of Neurology, Ibn Sina Hospital, POB 25427, Sabah Health Area, 13115, Kuwait; Email: emg.doctor@hotmail.com; Tel.: 00965 66 37 9970

Received: 25 Dec 2020 ♦ **Accepted:** 22 Mar 2021 ♦ **Published:** 30 June 2022

Citation: Alajmi A, Khuraibet A, Shamov T, Rousseff RT. Clinical-electrodiagnostic correlates in three cases of Riche-Cannieu anomaly (all-ulnar hand). Folia Med (Plovdiv) 2022;64(3):532-536. doi: 10.3897/folmed.64.e62461.

Abstract

The Riche-Cannieu anastomosis is a neural connection between the deep branch of the ulnar nerve and the recurrent motor branch of the median nerve to the thenar. Rarely, it provides complete or nearly complete ulnar innervation of the thenar muscles (all-ulnar hand). This may lead to confusing clinical and electrodiagnostic findings in patients with median or ulnar nerve pathology, or during workup for suspected nerve lesions.

Two patients referred for electrodiagnostic assessment of carpal tunnel syndrome and one patient with suspected peripheral neuropathy presented with absent or very low compound motor action potential from the thenar after median nerve stimulation. This occurred in spite of normal thenar muscles' bulk and function, in one hand (one subject) or both hands (two subjects). Ulnar nerve stimulation at wrist and elbow while recording over abductor pollicis brevis, first and second lumbrical muscles and needle electromyography allowed us to recognize Riche-Cannieu anastomosis and avoid diagnostic errors.

The rare innervation variant of all-ulnar hand should be considered in less typical presentations of hand disorders, especially when there is a discrepancy between the clinical findings and the expected electrodiagnostic changes.

Keywords

carpal tunnel syndrome, hand, median nerve, nerve conduction study, Riche-Cannieu anastomosis, ulnar nerve

INTRODUCTION

The textbook innervation of the thenar suggests the median nerve as predominantly supplying the abductor pollicis brevis, opponens pollicis, and the first and second lumbrical muscles. The flexor pollicis brevis muscle most often has dual median and ulnar supply.[1] However, significant variability of innervation exists in the hand, partially due to crossovers between the median and ulnar nerves.[2] The Riche-Cannieu anastomosis (RCA), named so after Riché and Cannieu who described it first, is a loop-shaped communication between the deep ulnar nerve branch and the recurrent thenar branch of the median nerve.[2,3] Recently, RCA is defined in the broader sense of ulnar-to-median motor connections at the hand, including patterns different from the classic thenar ansa. With microdissection, such branches are seen in almost all hands[3], but most of these seem to be of dubious significance because of their minute size, unclear direction, and fiber content.

Rarely, the ulnar nerve innervates entirely or almost entirely the intrinsic hand muscles. This extreme form of RCA is referred to as all-ulnar hand.[4,5] It results in con-

fusing clinical presentations and clinical-electrodiagnostic discrepancies and may cause diagnostic difficulties. The clinical manifestations of this innervation variant are mostly reported as single case reports or brief case series.[4,5]

CASE REPORT

This prospective case series was approved by the Hospital Ethics Committee and written consent was obtained from each patient. After recognition of the first case in January 2015, we were vigilant for this condition. Until April 2020, upper limb nerve conduction studies (NCS) were performed on 5428 patients at the Clinical Neurophysiology Unit which serves a population of about 560 000. During this period, we identified three cases of clinically significant RCA.

Case 1

A 48-year-old lady was referred for suspected right-sided carpal tunnel syndrome (CTS). She had painful nocturnal paresthesia in the right hand, with awakening and shaking the hand to alleviate the pain. Tinel and Phalen signs were positive on the right. Other history and the rest of her examination were non-contributory.

NCS and EMG were performed on a Nihon Kohden EMG machine (Neuropack MEB-9400) according to established protocols.[5] Unexpectedly, CMAP from the right APB was absent after median nerve stimulation at the wrist and elbow. Stimulation of the recurrent median motor branch in the palm produced a CMAP of normal size. Ulnar nerve stimulation at the wrist and elbow evoked an APB CMAP of similar size and shape. These findings, along with well-preserved thenar muscles bulk and strength, led to the suspicion for RCA. The CMAP from the first lumbrical muscle was absent with median nerve stimulation, but present within normal parameters after ulnar stimulation at the wrist and elbow. The right median antidromic sensory NCS and the palmar mixed comparison studies revealed mild abnormalities, supporting the clinical diagnosis of carpal tunnel syndrome.

Motor NCS on the left were normal and the median sensory studies - mildly abnormal, supporting asymptomatic median involvement.

The unusual findings suggesting RCA were underlined in the electrodiagnostic report. The patient's symptoms were managed by in the short term by wrist splinting. In a telephone interview at 6 months, she reported having a steroid injection with very good symptom control. Further follow-up was not available.

Case 2

A 44-year-old lady with unremarkable medical history presented with recent onset of paresthesia in the lateral three digits, wrist pain after driving, positive Phalen and Tinel signs on both sides. Muscle bulk and power in the hands were normal.

Wrist and elbow median nerve stimulation while recording from APB yielded low CMAP amplitudes of normal latency. Palmar stimulation of the median motor branch evoked normal CMAP bilaterally. Stimulation of the ulnar nerves at the wrist and the elbow produced normal CMAP with similar shape and size in APB. Lumbrical muscles 1 and 2 revealed a similar pattern of low CMAP after median nerve stimulation and a normal CMAP to ulnar stimulation at the wrist and elbow. Bilaterally, needle EMG examination of APB was normal. The median sensory NCS were mildly abnormal on both sides, supporting the clinically suspected mild CTS.

The patient was referred to an orthopedist, noting the innervation variant. At the telephone interviews at 3 and 6 months, she reported effective control by splinting without further interventions.

Case 3

A 30-year-old lady with an unrevealing medical history and exam was referred to us to "exclude peripheral neuropathy" because of transient glove-stoking paresthesias. Her NCS revealed low CMAP with normal latency from APB, LM1 and LM2 on both sides after median nerve stimulation, but normal size and shape after ulnar stimulation at the wrist and elbow. Sensory NCS were normal. The needle EMG of both APB muscles was also normal. She was referred back to her general practitioner to rule out electrolyte disturbances or hyperventilation syndrome. We have no follow-up for this patient.

Selected graphs of patients' NCS are presented in ***Figs 1, 2, 3****.*

DISCUSSION

While the incidence of frequency of Riche-Cannieu anastomosis or anomaly varies widely according to its definition, the all-ulnar hand is a rarity.[4,5] In the neurologically healthy person, it is asymptomatic and may be discovered incidentally in subjects referred for NCS for symptoms unrelated to median/ulnar pathology.[6]

In median neuropathy below the elbow (most frequently CTS), the RCA may prove misleading in two ways. In extreme cases of CTS or complete median transection, the clinically preserved thenar function via the anastomosis may mask the real severity of the lesion; out of 101 CTS patients with absent CMAP, over 10% had mild or no muscle atrophy and intact thumb opposition and could be considered mild on purely clinical grounds.[7] Indeed, many hand surgeons tend to neglect the NCS and emphasise on thenar atrophy as a main decision-making factor in CTS workup.[8] The reverse is also possible: over-relying on the electrodiagnostic absence of thenar CMAP may lead to an erroneous diagnosis of "extreme" CTS while the neuropathy is mild or even absent.[9]

With ulnar nerve lesions, RCA is reported as casuistic in single patients but is even more deceptive. Deep ulnar branch lesion in the hand (posttraumatic, piso-hamate hi-

Folia Medica

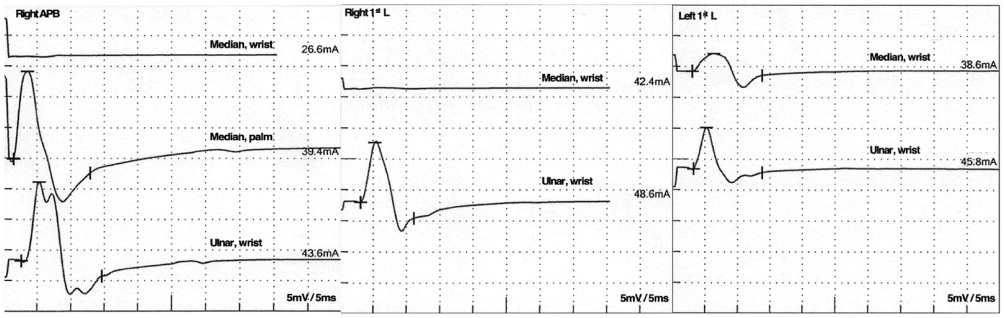

Figure 1. Patient 1. The absence of CMAP from APB and first lumbrical muscles after median nerve stimulation at the wrist is evident. Responses appear after ulnar stimulation. On the left, conduction to the first lumbrical muscle is normal (presented for comparison).

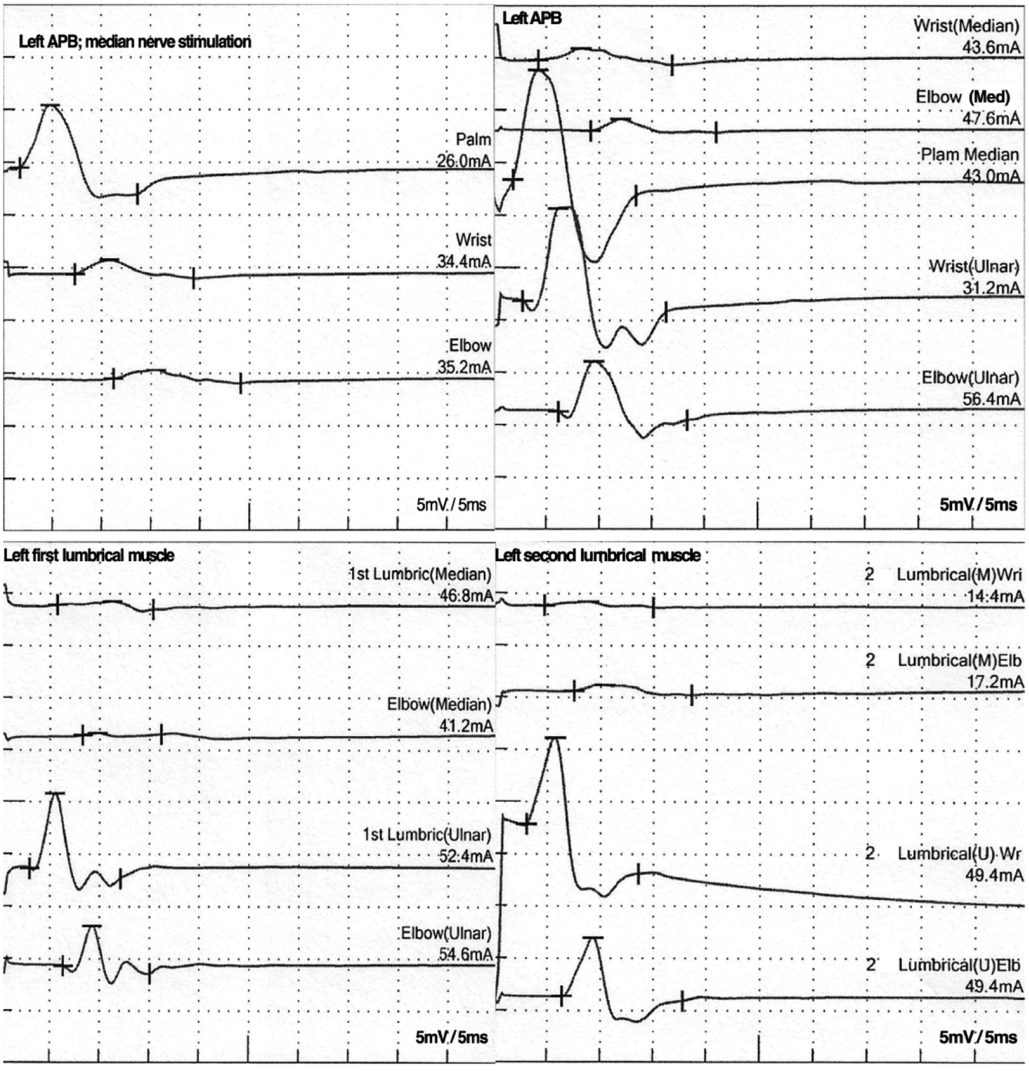

Figure 2. Patient 2. Very low response from the APB, first and second lumbrical muscles after median stimulation, high CMAPs after ulnar stimulation.

atus syndrome) leads to painless atrophy of both the thenar and the interossei muscles, imitating the "split hand atrophy" of motor neuron disease.[10,11] With the ominous prognosis it carries, such misdiagnosis becomes very burdensome. In proximal ulnar lesions, the manifest thenar involvement may falsely suggest plexus or root lesion and cause unnecessary investigations and delay in treatment.[12]

In such conflicting clinical-electrodiagnostic situations,

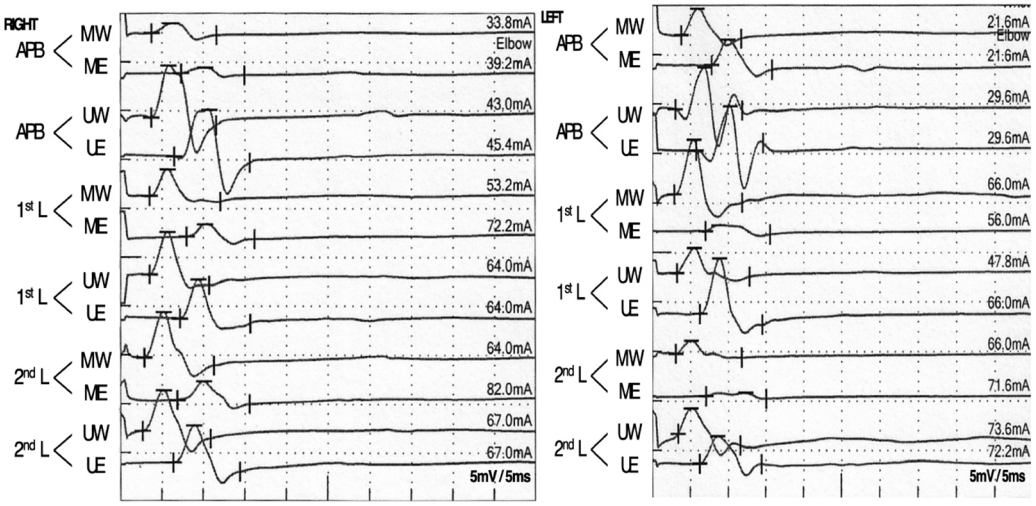

Figure 3. Patient 3. Findings similar to those in patient 2, results for both upper limbs are presented.

one should compare the median and ulnar conduction to the thenar after stimulation at the wrist and elbow, strictly avoiding the possibility of co-stimulation of both nerves or co-registration of CMAP from different muscles.[4,5] This may require needle recording and near-nerve needle stimulation. Recording from the lumbrical muscles and needle EMG study of thenar muscles are also needed. Recently, MRI or ultrasound imaging of the deep ulnar branch in the hand has been applied in diagnosis.[11]

CONCLUSIONS

In case of apparent clinical and electrodiagnostic discrepancies during workup of suspected median and/or ulnar nerve pathology, the rare all-ulnar hand innervation variant (an extreme form of RCA) should be considered to avoid possible diagnostic errors and delay.

Disclosures

All authors fulfil the authorship criteria by their contributions to data acquisition, designing, drafting, editing, and approving the paper. The authors declare no conflict of interest. No funding was received for the production of this manuscript.

REFERENCES

1. Ross AC. Wrist and hand. In: Standring S, ed. Gray's Anatomy, 41st ed. New York: Elsevier Limited; 2016: 886–7, 891–3.
2. Yang H, Gil Y, Kim S, et al. From the brachial plexus to the hand, multiple connections between the median and ulnar nerves may serve as bypass routes for nerve fibres. J Hand Surg Eur Vol 2016; 41:648–56.
3. Caetano E, Vieira L, Sabongi Neto J, et al. Riché-Cannieu anastomosis: structure, function, and clinical significance. Rev Bras Ortop (Sao Paulo) 2019: 54:564–71.
4. Oh S. Clinical electromyography: nerve conduction studies. 3rd ed.; Philadelphia PA, USA: Lippinkot Williams & Wilkins; 2003; 356–8.
5. Van Dijk JG. Anomalies of innervation. In: Kimura J, Daube JR, Mauguière F, editors. Handbook of clinical neurophysiology. Amsterdam: Elsevier; 2006; Vol. 7:311–33.
6. Boland R, Krishnan A, Kiernan M. Riche-Cannieu anastomosis as an inherited trait. Clin Neurophysiol 2007; 118:770–5.
7. Ebata T, Imai K, Tokunaga S, et al. Thumb opposition in severe carpal tunnel syndrome with undetectable APB-CMAP. Hand Surg 2014; 19:199–204.
8. Fowler J. Nerve conduction studies for carpal tunnel syndrome: gold standard or unnecessary evil? Orthopedics 2017; 40:141–2.
9. Hess C, Jenkins J. Riche-Cannieu anastomosis masquerading as severe median neuropathy: a case report. PM&R 2012; 10:S229–30.
10. Saperstein D, King R. Motor neuron presentation of an ulnar neuropathy and Riche-Cannieu anastomosis. Electromyogr Clin Neurophysiol 2000; 40:119–22.
11. Rovers J, Brekelmans G, Visser L. Piso-hamate hiatus syndrome in a patient with Riche-Cannieu anastomosis. BMJ Case Rep 2013; 2013:bcr2013008601.
12. Dumitru D, Walsh NE, Weber CF. Electrophysiologic study of the Riche-Cannieu anomaly. Electromyogr Clin Neurophysiol 1988; 28:27–31.

Folia Medica

Клинико-электродиагностические корреляты в трёх случаях анастомоза Рише-Канью (типичная „локтевая“ кисть)

Абдулла Аладжми[1], Аднан Кураибет[1], Тодор Шамов[2], Росен Русев[1,3]

[1] *Отделение неврологии, Больница „Ибн Сина“, Кувейт*

[2] *Отделение нейрохирургии, Военно-медицинская академия, София, Болгария*

[3] *Кафедра неврологии и нейрохирургии, Плевенский медицинский университет, Плевен, Болгария*

Адрес для корреспонденции: Росен Русев, Отделение неврологии, Больница „Ибн Сина“, Медицинский район „Саба“, POB 25427, 13115, Кувейт; Email: emg.doctor@hotmail.com; Тел.: 00965 66 37 9970

Дата получения: 25 декабря 2020 ♦ **Дата приемки:** 22 марта 2021 ♦ **Дата публикации:** 30 июня 2022

Образец цитирования: Alajmi A, Khuraibet A, Shamov T, Rousseff RT. Clinical-electrodiagnostic correlates in three cases of Riche-Cannieu anomaly (all-ulnar hand). Folia Med (Plovdiv) 2022;64(3):532-536. doi: 10.3897/folmed.64.e62461.

Резюме

Анастомоз Рише-Канью представляет собой нервное соединение между глубокой ветвью локтевого нерва и возвратной двигательной ветвью срединного нерва к тенару. В редких случаях он обеспечивает полную или почти полную локтевую иннервацию мышц тенара (типичная „локтевая“ кисть). Это может привести к путанице в клинических и электродиагностических данных у пациентов с патологией срединного или локтевого нерва или во время обследования при подозрении на поражение нерва.

Два пациента были направлены на электродиагностическую оценку синдрома запястного канала, и один пациент с подозрением на периферическую невропатию имел отсутствующий или очень низкий составной двигательный потенциал тенара после стимуляции срединного нерва. Это произошло, несмотря на нормальную массу и функцию мышц тенара, в одной руке (один субъект) или обеих руках (два субъекта). Стимуляция локтевого нерва на уровне запястья и локтя с записью над короткой отводящей большой палец, первой и второй червеобразными мышцами и игольчатая электромиография позволили распознать анастомоз Рише-Канью и избежать диагностических ошибок.

Редкий вариант иннервации типичной „локтевой“ кисти следует рассматривать при менее типичных проявлениях нарушений кисти, особенно когда есть несоответствие между клиническими данными и ожидаемыми электродиагностическими изменениями.

Ключевые слова

синдром запястного канала, кисть, срединный нерв, исследование нервной проводимости, анастомоз Рише-Канью, локтевой нерв