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# Combined Central and Peripheral Demyelination with a Favorable Outcome: A Case with 10-Year Follow-up

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## ABSTRACT

**Introduction:** Combined central and peripheral demyelination (CCPD) is a rare and heterogeneous condition, often associated with a poor prognosis. Contrary to the popular reports, we present a case of CCPD with a favorable prognosis and a 10-year follow-up.

**Case:** A 29-year-old female patient presented with progressive limb weakness following a Brucella infection. She was subsequently diagnosed with motor type chronic inflammatory demyelinating polyneuropathy (CIDP), which was characterized by motor conduction blocks and demyelinating motor conduction abnormalities in a nerve conduction study (NCS). In the following years, the patient was diagnosed with CCPD with relapse and remissions, with or without CIDP attacks and multiple cranial neuropathy, cervical myelitis, and optic neuritis attacks. Despite exhibiting motor CIDP, she did not respond to intravenous immunoglobulin (IVIg) therapy, and all her episodes were removed with intravenous methylprednisolone (IVMP).

**Conclusion:** Our case has been in remission with azathioprine therapy for the last three years and is notable for its prolonged course, good prognosis, and potential association with Brucella infection.

**Keywords:** Brucella, cervical myelitis, motor chronic inflammatory demyelinating polyneuropathy, multiple cranial neuropathy.

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## INTRODUCTION

Combined central and peripheral demyelination (CCPD) is a clinical condition with a rare and heterogeneous clinical presentation (1)**.** It usually has a poor prognosis, with patients commonly presenting as acute, relapsing-remitting, or chronic forms. Clinical follow-up often shows either simultaneous or sequential involvement of central and peripheral systems (1–3). Here we present a case of relapsing-remitting CCPD with varied clinical presentations and, contrary to typical reports, an unexpectedly favorable long-term prognosis.

## CASE

A 29-year-old female patient was admitted to the neurology outpatient clinic with progressive weakness of the limbs and mild sensory symptoms for approximately one year, in 2014. A year ago, she was treated for brucellosis and the neurological symptoms began following the illness. The neurological examination showed intact cranial nerve function, 4/5 muscular strength in both the proximal and distal limbs, and global areflexia. Electromyography was performed, which showed motor conduction blocks while sensory conduction in the upper and lower extremities was normal (Table 1), suggestive of motor-type peripheral neuropathy with demyelinating features (motor CIDP). In this period, intravenous immunoglobulin (IVIg) treatment was administered for 5 days, followed by monthly IVIg and her clinical status did not improve. Therefore, the treatment was switched to intravenous methylprednisolone (IVMP) therapy and the patient's clinical status improved. The patient continued to receive monthly IVMP treatment during this period, her neurological examination returned to normal, and the control NCS improved. Remission was achieved.

One year later from the remission, weakness in the proximal lower extremities recurred, and motor-type demyelinating peripheral neuropathy with symmetrical conduction blocks was observed again on NCS. The lumbar puncture showed increased CSF protein levels with albuminocytological dissociation. Simultaneous oligoclonal band type 1 was negative and Ig G index was within the normal limit. Ganglioside panel was negative. The serum sample tested positive for Brucella Coombs at a titer of 1/160, whereas the CSF sample tested negative. Vasculitis markers were negative. Intravenous methylprednisolone treatment was administered and the neurological condition improved. The NCS conducted in the following year showed no abnormalities (Table 2).

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## Highlights

- **The case of CCPD with a good prognosis following brucella infection is presented.**
- **Unlike most cases of CCPD, the attacks were resolved without sequelae with MP treatment.**
- **Although not the first choice for treatment, remission was achieved with AZA for 3 years.**

In 2017, she was presented with bilateral 6th cranial nerve paresis and facial diplegia. The NCS results were within normal limits, except for the observed prolonging of F latencies. Brain MRI was normal. In this period, IV methylprednisolone 1 g/day was administered for 8 days, followed by oral prednisolone 1 mg/kg/day (total 80 mg/day), and her clinical status improved.

In 2019, she presented sudden imbalance and left arm weakness. Cervical MRI showed a demyelinating lesion at the level of C3–4 in a vertical segment of approximately 12–13 mm, involving the central part and the left half of the spinal cord (Fig. 1). Nerve conduction study was observed demyelinating peripheral neuropathy with motor conduction blocks in the bilateral median and ulnar nerves. Anti-myelin oligodendrocyte glycoprotein (anti-MOG) and Aquaporin 4 (AQP4) antibodies in serum were negative. Clinical improvement was observed with 5 days of IV methylprednisolone.

In 2020, she developed left peripheral facial paralysis was treated with pulse steroid therapy and recovered without sequelae. In 2021, the patient developed decreased visual acuity in the left eye, and VEP examination showed delayed P100 latency on the left. 5 days IVMP treatment was given in this period due to optic neuritis. The patient's complaints completely regressed. Azathioprine was added to the patient's treatment.

In the follow-up, low-dose prednisolone was added to AZA treatment upon the detection of demyelinating neuropathy with motor conduction block in the NCS performed in 2024. During this period, neurofascin 155 and contactin, Caspr1 were found to be negative while under immunosuppressive treatment. Three months later, the NCS returned to normal.

Since 2021, the patient has had no neurological complaints and the neurological examination has been normal.

**Table 1.** Nerve conduction study at initial arrival

| Nerve | Stimulation site | Recording site | Amplitude (mV) | Latency (ms) | Conduction velocity (m/s) | F wave (ms) |
|---|---|---|---|---|---|---|
| **Motor** | | | | | | |
| **Median, R** | Wrist | APB | 10.8 | 3.15 | | |
| | Elbow | APB | 6.9 | 9.20 | 36.4 | 47.2 |
| | Axilla | APB | 2.7 | 16.40 | 30.6 | |
| **Median, L** | Wrist | APB | 10.5 | 3.15 | | |
| | Elbow | APB | 3.2 | 10.25 | 35.2 | 41.1 |
| | Axilla | APB | 1.4 | 18.80 | 32.4 | |
| **Ulnar, R** | Wrist | ADM | 11.0 | 3.10 | | |
| | B. Elbow | ADM | 6.5 | 8.85 | 41.3 | 48.8 |
| | A. Elbow | ADM | 4.6 | 11.55 | 33.3 | |
| | Axilla | ADM | 3.5 | 22.60 | 19.5 | |
| **Ulnar, L** | Wrist | ADM | 12.7 | 3.00 | | |
| | B. Elbow | ADM | 3.2 | 8.85 | 44.2 | 44.6 |
| | A. Elbow | ADM | 2.2 | 11.30 | 36.7 | |
| | Axilla | ADM | 1.1 | 21.70 | 22.6 | |
| **Tibial , L** | Ankle | AH | 11.8 | 5.35 | | 77.4 |
| | Popliteal | AH | 2.0 | 15.15 | 34.8 | |
| **Tibial , R** | Ankle | AH | 15.1 | 4.75 | | 69.5 |
| | Popliteal | AH | 4.1 | 14.40 | 33.5 | |
| **Peroneal, R** | Ankle | EDB | 4.1 | 5.5 | 28.8 | 78.1 |
| | B. Fibula | EDB | 3.1 | 18.95 | | |
| **Peroneal, L** | Ankle | EDB | 3.5 | 5.65 | 28.4 | 75.8 |
| | B. Fibula | EDB | 2.0 | 17.75 | | |
| **SENSORY** | | | | | | |
| **Median, L** | Wrist | Second Finger | 12.9 | 2.40 | 58.3 | |
| | Elbow | Wrist | 42.9 | 1.15 | 69.6 | |
| **Median, R** | Wrist | Second Finger | 11.4 | 2.55 | 54.9 | |
| | Elbow | Wrist | 50.5 | 1.25 | 64.0 | |
| **Ulnar, L** | Wrist | Fifth Finger | 8.8 | 2.35 | 59.6 | |
| **Ulnar, R** | Wrist | Fifth Finger | 8.6 | 2.45 | 57.1 | |
| **Sural, R** | Calf | Lateral Malleol | 14.4 | 2.1 | 42.9 | |
| **Sural, L** | Calf | Lateral Malleol | 15.6 | 2.3 | 42.6 | |

Note that all nerves in the motor conduction studies exhibit conduction block, F-wave latencies are prolonged within demyelinating ranges, and motor conduction velocities are slowed, while sensory conduction studies remain normal. Amplitudes are measured in millivolt (mV, motor) and in microvolt (μV, sensory). APB: abductor pollicis brevis; ADM: abductor digiti minimi; EDB: extensor digitorum brevis; AH: adductor hallucis; R: right; L: left.

**Table 2.** Re-examined nerve conduction study of right extremities after treatment

| | Stimulation site | Recording Site | Amplitude | Latency (ms) | Conduction Velocity (m/s) | F wave (ms) |
|---|---|---|---|---|---|---|
| **Motor** | | | | | | |
| **Median** | Wrist | APB | 10.9 | 3.05 | | 28.30 |
| | Elbow | APB | 8.5 | 7.35 | 50.5 | |
| **Ulnar** | Wrist | ADM | 9.5 | 2.95 | | 31.50 |
| | B. Elbow | ADM | 7.5 | 5.90 | 62.7 | |
| | A. Elbow | ADM | 7.2 | 7.80 | 47.4 | |
| **Tibial** | Ankle | AH | 13.7 | 4.10 | | 48.10 |
| | Popliteal | AH | 9.5 | 12.85 | 43.4 | |
| **Peroneal** | Ankle | EDB | 3.6 | 4.15 | | 51.2 |
| | B. fibula | EDB | 3.0 | 10.40 | 48.0 | |
| | Popliteal | EDB | 3.0 | 11.90 | 53.3 | |
| **SENSORY** | | | | | | |
| **Median** | Wrist | Second finger | 16.4 | 2.60 | 59.6 | |
| **Ulnar** | Wrist | Fifth finger | 7.3 | 2.3 | 60.9 | |
| **Sural** | Calf | Lat malleol | 13.50 | 2.05 | 39.0 | |

Amplitudes are measured in millivolt (mV, motor) and in microvolt (μV, sensory). APB: abductor pollicis brevis; ADM: abductor digiti minimi; EDB: extensor digitorum brevis; AH: adductor hallucis.

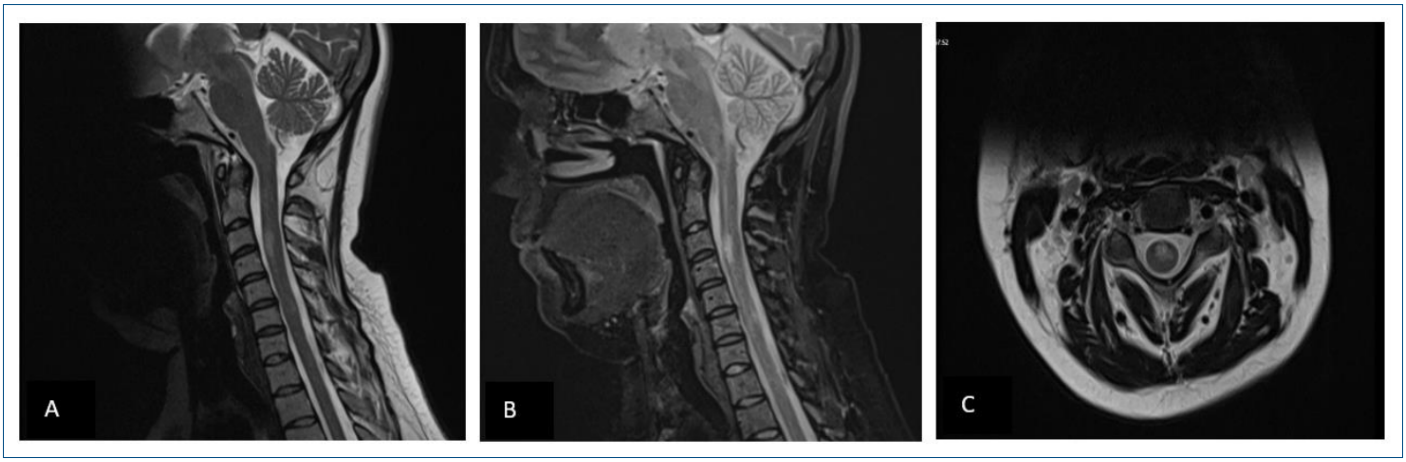


**Figure 1.** Cervical MRI images showed short a strip and high-intensity signals from 3rd to 4th cervical level. (A) T1WI; (B) T2WI; (C) the horizontal cross-section of 3rd cervical level (MRI: magnetic resonance imaging; T1WI: T1-weighted images; T2WI: T2- weighted images).

## DISCUSSION

We present a 10-year case study of a young woman diagnosed with CCPD, who initially exhibited motor Chronic Inflammatory Demyelinating Polyneuropathy (CIDP), followed by multiple episodes of cranial neuropathy, cervical myelitis, and optic neuritis, ultimately resulting in favorable prognosis. In contrast with the literature, this case is remarkable due to its long follow-up period, steroid responsiveness despite having a predominant clinical course with motor CIDP and being in remission with azathioprine for three years.

Considering the patient's initial complaints and electrophysiological examinations, the motor variant appears compatible with CIDP according to the European Federation of Neurological Societies/Peripheral Nerve Society 2021 diagnostic criteria (4). Since motor CIDP treatment patients may worsen with steroids, IVIg is recommended as the first option in such cases (4). However, in our case, there was no response to IVIg. In this case, autoimmune nodopathy could be compatible due to the young age of onset, relatively low response to IVIg treatment, cranial neuropathy attacks and ataxia in one of the attacks (3–5). However, neurofascin 155/186 and contactin1/caspr1 were found to be negative.

Our case does not fulfill the McDonald 2017 Multiple Sclerosis diagnostic criteria's spatial spread criteria for central involvement. While optic neuritis and cervical demyelinating lesion suggest a typical clinic for anti-MOG-associated disease (6), the anti-MOG antibody test, performed using the ELISA method, was negative. Furthermore, these clinical symptoms could be correlated with neuromyelitis optica spectrum disorder (NMOSD). However, anti AQP4 antibody test was negative. On the other hand, the lack of gadolinium-enhancement T1 lesion involving more than half of the optic nerve or T2 hyperintensity in the optic neuritis and the short segment of the myelitis attack do not fulfill the criteria for seronegative NMOSD (7). In the literature, CCPD cases that do not meet these criteria have also been described (1,3,8).

Combined central and peripheral demyelination may be observed following an antecedent infection (1–3). The fact that the patient's complaints started after brucella infection in the first attack and serum brucella was found to be positive suggests that it may develop secondary to brucella infection. There have been reports of cases of axonal neuropathy and CIDP associated with Brucella infection (9–10). Guillian Barre Syndrome has rarely been reported (11). Our case is the first to

suggest that brucella could be a prodromal factor for CCPD, beyond the infections previously described in the literature.

Combined central and peripheral demyelination case series indicates that pulse steroid therapy is more beneficial particularly during the acute stage (1–3). In the follow-up of the disease, rituximab may be effective in addition to other treatments (1). Nevertheless, the majority of the cases seem to result in sequelae or display no improvement in modified Rankin scores.

There are cases with good prognosis in the literature (12,13). However, no patient similar to our case has been reported to have good prognosis with AZA and steroid treatment. Although we could not conclusively prove the etiology of CCDP or associated antibodies. Our case is a unique example of CCPD having a motor CIDP component with a good prognosis, as opposed to what is typically documented in the literature, resulting in a complete recovery with steroid and azathioprine therapy. Good prognosis can be achieved with appropriate immunosuppressive treatment combinations. Further studies are needed to identify the clinical, neuroradiological, and electrophysiological factors associated with favorable outcomes.

**Informed Consent:** Ethics approval Informed consent was obtained from the patient for case description and publication.

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**Conflict of Interest:** The authors declared that there is no conflict of interest.

**Financial Disclosure:** None.

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