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Case Report

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Autoimmune hepatitis complicated with entrapment neuropathy & fibromyalgia: A case report

Abstract

46. y-old right-handed female with history of controlled autoimmune hepatitis since 2001 with negative liver biopsy on 2017 presented with chronic generalized pain affecting neck, upper back, buttocks, knee and chest associated with paresthesia and tingling of left hand with nocturnal exacerbation. Clinical exam revealed; Neurologically Positive Tinel sign over median nerve at wrist bilaterally, Positive Phalen test bilaterally, Musculoskeletal exam revealed multiple tender points bilaterally with limited neck Rom due to pain. Electrophysiological testing of both upper extremities documented bilateral median sensory neuropathy at wrist, demyelinating pathology and bilateral ulnar nerve entrapment at wrist: Guyon canal syndrome; sensory-motor axonal demyelinating. Normal liver and kidney function tests and glycosylated hemoglobin 5.

Keywords: autoimmune hepatitis, fibromyalgia, entrapment neuropathy.

Volume 7 Issue 2 - 2022

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Received: August 22, 2022 | **Published:** September 15, 2022

Introduction

Neurological manifestations have been identified in the context of autoimmune hepatitis (AIH). [1]Previous case reports highlighted the association between AIH and sensory neuronopathy (SN). Despite that, little is known about the frequency of AIH-related SN and its clinical/neurophysiological profile.[1] Moreover, it is not clear whether SN is an AIH-specific manifestation or related to chronic liver damage, SN was identified only in AIH patients (5/70 vs. 0/52, P=0.04); the overall prevalence of AIH-related SN was 7% with an average profile of a woman in her 40s with asymmetric onset of sensory deficits that chronically evolved to disabling proprioceptive ataxia associated with marked dysautonomia. Neurological disability and hepatocellular damage did not follow in parallel. Anti-fibroblast growth factor receptor type 3 antibodies were found in 3/5 (60%) of the patients with AIH-related SN.[1] Fibromyalgia was found in association with autoimmune diseases.[2]

Case report

46 y-old Right-handed females with history of controlled autoimmune hepatitis since 2001, treated with azathioprine &steroids and has negative liver biopsy (2017), presented with chronic generalized pain affecting neck, upper back, chest, both knees for many years in addition to numbness of 4th &5th left digits and tingling and paresthesia of left hand with nocturnal exacerbation. Pain by visual analogue scale 9-10/10. Clinical examination; Musculoskeletal exam: Neck: limited ROM due to pain, Negative Spurling test bilaterally. Multiple tender points over both trapezii, latissimus dorsi, rhomboids, supraspinatous, infraspinatous, buttocks, lateral epicondyles, medial aspect of both knees, 2nd costo-chondral junctions. Neurological exam: Deep tendon jerks of both upper extremities G2, Sensation of both upper extremities: Intact. Manual muscle testing of both upper extremities muscles: full. positive Tinel signs for both median nerves at wrist and Positive Phalen's test, bilaterally. Electrophysiological testing revealed delayed median sensory peak latency bilateral, delayed ulnar peak sensory and motor latency with diminished ulnar CMAP and normal conduction across dorsal cutaneous nerve. Normal electromyography of both upper extremities suggesting bilateral sensory demyelinating median neuropathy at wrist & bilateral sensory -motor axonal demyelinating ulnar neuropathy at wrist; Guyon canal syndrome. Normal liver, kidney, rheumatological testing, HgA1c=5.

Discussion

A patient with controlled autoimmune hepatitis recently suffer from entrapment neuropathy with No diabetes that could explain the condition. In addition to generalized pain fulfilling fibromyalgia criteria.

Martinez ARM et al.[1] reported prevalence of around 7% of sensory polyneuropathy mainly sensory ataxia in Autoimmune hepatitis patients not parallel to liver damage.[1] But this patient has entrapment neuropathy only.

Park KH et al.[3] suggested that Sensory neuropathy was rarely encountered with autoimmune hepatitis and could be immune mediated.

They reported that AIH related peripheral neuropathy has been documented with various clinical presentations,[2] which include chronic inflammatory demyelinating polyneuropathy,[4] mononeuritis multiplex,[5] motor -axonal polyneuropathy,[6] sensory neuropathy.[7] Those AIH related neuropathy reported dramatic improvement by intravenous immunoglobulin but not to steroid.

Chaudhry V et al.[8] reported axonal polyneuropathy with end stage liver disease with different etiologies. In the contrary, this case, had normal liver function and neuropathy is mainly demyelinating entrapment neuropathy

The above-mentioned study did not suggest the entrapment neuropathy as a common feature of liver disease, moreover, this particular case had controlled autoimmune hepatitis with negative enzymes and biopsy for years i.e., liver return to normal, not end stage and still associated with fulminant entrapment neuropathy and fibromyalgia.

The suggested mechanism of neuropathy is immune mediated. Cojocaru IM et al.[9] suggested that hepatitis B &C associated with polyarteritis nodosa with immune complexes to viral protein detected in those patients with mononeuritis multiplex or symmetrical neuropathy.[9] These findings were associated with abnormal or sometimes normal liver function. Cojocaru IM et al also suggested that hepatitis associated with mixed cryoglobulinemia or associated with vasculitis.[9]

Schnedl W et al.[10] suggested that the pathogenesis of neuropathy of AIH is not known but may stem from deposit of immune complexes in vasa nervosum which then cause vasculitis and ischemia of nervous fibers as was shown with cryoglobulinemia

Fibromyalgia was reported in autoimmune diseases. Chronic pain leads to central sensitization and in chronic liver disease with chronic abdominal pain, it could be the reason of central sensitization, hence Fibromyalgia.[2]

Rogal SS et al.[11] suggested fibromyalgia symptoms were significantly associated with hepatitis C virus and non- alcoholic steato-hepatitis cirrhosis.[11]

Rivera J et al.[12] suggested that fibromyalgia is associated with Hepatitis C infection, but not necessarily associated with liver damage or elevated liver damage12. In addition, the presence of fibromyalgia was not associated with elevated immune markers. The proposed explanation how infection can trigger fibromyalgia- first direct infection of host tissues or inflammatory mediators released during infection could induce fibromyalgia. Some cytokines also suggested. Stress & anxiety are also suggested mechanisms. No clear association between severity of HCV & FM. Age & female sex are important factors in appearance of FM.

Mohamed RHA et al.[13] studied the prevalence of rheumatological manifestations among Egyptian chronic hepatitis C patients, reported chronic fatigue syndrome (9.5%), Sicca symptoms (8.8%), arthralgia (6.5%), fibromyalgia 1.9%, myalgia, thrombocytopenia 0.7%, cryoglobulinemia, vasculitis 0.7%. The fibromyalgia was mainly presented in female populations.[13]

Conclusion

Autoimmune hepatitis, although controlled for many years, can still be complicated with entrapment neuropathies as well as fibromyalgia. The typical neuropathy listed in the literature was sensory polyneuropathy. And the suggested mechanism was Immune mediated.

Acknowledgments

None.

Funding

None.

Conflicts of interest

Author declares there is no conflict of interest exists.

References

1. Martinez ARM, De Lima FD, Martins MP, et al. Sensory neuronopathy is a specific and disabling neurological manifestation of autoimmune hepatitis. *European journal of neurology*. 2020;27(10):2072–2078.
2. Xavier de Oliveria DM, Uchoa da Silva RDS. Rheumatological manifestations associated with viral hepatitis B or C. *Rev Soc Bras Med Trop*. 2019;52:e20180407.
3. Park KH, Sohn SY, Sung JJ, et al. Subacute sensorimotor polyneuropathy associated with autoimmune hepatitis. *Journal of Clinical Neurology*. 2016;12(1):123–125.
4. Domingos JP, Garrido C, Moreira Silva H, et al. Chronic inflammatory demyelinating polyneuropathy associated with autoimmune hepatitis. *Pediatr Neurol*. 2014;51(3):e13–e14.
5. Luth S, Birklein F, Schramm C, et al. Multiplex neuritis in a patient with autoimmune hepatitis: a case report. *World J Gastroenterol*. 2006;12(33):5396–5398.
6. Schnedl WJ, Krause R, Tafeit E,et al. Autoimmune hepatitis associated with motor-axonal polyneuropathy. *Digestion*. 2009;80:50–51.
7. Merchut MP, Adams EM, Morrissey M. Sensory neuronopathy in autoimmune chronic active hepatitis. *Neurology*. 1993;43(11):2410–2411.
8. Chaudhry V, Corse Am, O'brian R,David R, et al. Autonomic and peripheral (sensorimotor) neuropathy in chronic liver disease: a clinical and electrophysiologic study, peripheral neuropathy. 2nd edn. Philadelphia: Saunders; 1984:1826–1832.
9. Cojocaru,Im, Cojocaru M, Silosi I, et al. Peripheral nervous system manifestations in systemic autoimmune diseases. *Maedica (Bucur)*. 2014;9(3):289–294.
10. Rogal SS, Bielefeldt K, Di Martini AF, et al. Fibromyalgia symptoms & cirrhosis. *Dig Dis Sci*. 2015;60(5):1482–1489.
11. Riera J, Diego AD, Trinchet M, et al. Fibromyalgia associated hepatitis C virus infection. *Br J Rheumatic.* 1997;36(9):981–985.
12. Mohamed RHA, Elmakhzany HI, Gamal A, et al. Prevalence of rheumatologic manifestations of chronic hepatitis C virus infection among Egyptians. *Clin Rheumatol*. 2010;29(12):1373–1380.