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Case study

An uncommon complication of *Listeria monocytogenes* infection: Polyradiculoneuritis following Listeria meningoencephalitis

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ABSTRACT

Listeria monocytogenes, primarily a foodborne pathogen, is commonly responsible for disorders affecting the central nervous system and cranial nerves. We hereby present the first case to our knowledge of listeriosis linked to a peripheral neurological disorder causing acute upper limb weakness.

Introduction

Listeria monocytogenes is a saprophytic Gram-positive aerobic bacterium, ubiquitously found in nature that more generally affects neonates, older adults, immunosuppressed patients or pregnant women. Nevertheless, some cases occur in healthy population [1]. Listeriosis is generally an illness due to consumption of contaminated food including unpasteurized milk or cheese as in the 1987 Swiss epidemic due to Vacherin Mont d'Or cheese [2]. Some direct transmission from animals and vertical transmission have also been described [1]. Although *Listeria monocytogenes* is responsible for many different neurological manifestations such as rhomboencephalitis, cerebritis, or focal neurological deficits such as aphasia, hemiparesis and cranial nerve palsies, acute meningoencephalitis remains one of the most common forms of listeriosis in adults with clinical features such as mental status change, fever, headache, tremor, seizures, and coma [1,3].

To our knowledge, no peripheral neurological complication such as polyradiculoneuritis has yet been described. We report below a case of *Listeria monocytogenes* meningoencephalitis with a para-infectious acute upper limb weakness.

Case presentation

A 79-year-old Caucasian woman developed high fever (up to 40 °C), headache, nausea and vomiting over 2 days. She came to attention after presenting a mild confusional state. In the emergency room, she had a body temperature of 39 °C, a respiratory rate of 24/min, normal pulse and an elevated blood pressure at 170/70 mm Hg. She was alert and orientated but had slightly reduced mental speed and some difficulty completing complex tasks. The neck was stiff with positive Brudzinski and Kernig signs but no other neurological abnormality on examination. The lumbar puncture (LP) revealed elevated leukocytes (624/μL with 21% of polynuclear cells and 79% of mononuclear cells), elevated protein (1683 mg/L) and normal glucose. *Listeria monocytogenes* was identified by PCR assay from microbiological analysis of cerebrospinal fluid. Blood cultures were sterile. Her past medical history was relevant for renal and bone tuberculosis at the age of 15 and hemochromatosis with a heterozygote HFE mutation. Diagnosis of meningoencephalitis due to *Listeria monocytogenes* was made leading to a treatment of amoxicillin and gentamicin for 2 weeks and amoxicillin alone for 2 more weeks with a rapid and favorable response. Further information revealed that the patient had consumed unpasteurized goat cheese and raw milk a few days before the first symptoms.

Three days after admission, the patient presented a progressive loss of strength beginning in her right arm. The neurological exam showed moderate and diffuse weakness of the upper limbs (right more than left), predominant in the distribution of the extensor muscles of the wrist and fingers where gravity could not be overcome (MRC scale 3). There was no sensation or proprioception alteration and tendon reflexes were preserved. The patient noticed at the same time a slight change in taste. Apart from this, all cranial nerves were intact. ECG monitoring revealed an asymptomatic transient supraventricular tachycardia. The work up included cerebral and cervical spine MRI showing no abnormality and no contrast enhancement of the cervical roots.

A nerve conduction study at day 6 after admission found an inhomogeneous acute axonal more than demyelinating peripheral nerve disorder affecting only the upper arms and sparing the sensory nerves. The motor conduction studies were normal for the median and ulnar

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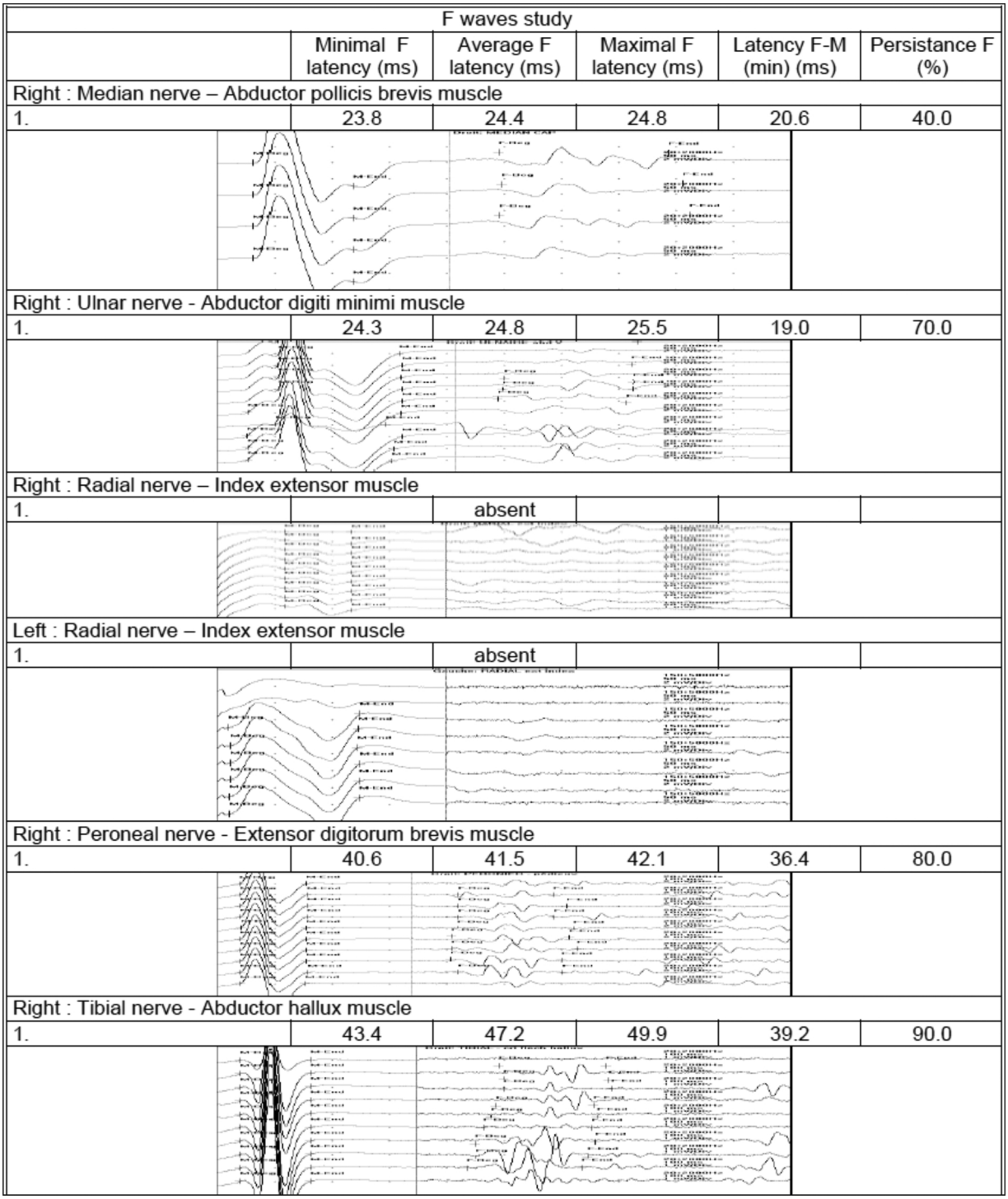

Fig. 1. F waves study. Right median, ulnar and radial nerves. Left radial nerve. Right peronea l and tibial nerve.

nerves, but showed, for the radial nerve, decreased distal amplitude and partial conduction block with proximal stimulation (shown by the further loss of amplitude and mild reduction of velocity). F waves reflecting the radial proximal responses were bilaterally absent while F waves of median and ulnar nerves were normal. [Fig. 1]. Needle examination demonstrated poor recruitment in the index extensor muscle with no change in morphology of muscle unit potentials. We considered the diagnosis of acute polyradiculoneuritis but did not decide to administer intravenous immunoglobulin given the non-disabling deficits.

Screening for *Mycobacterium Tuberculosis* with IFN gamma release assay and HIV serology were normal as vitamin levels (B6, B9 and B12). Anti-HEV IgM antibodies were elevated (1.9 for a normal range < 1.0) despite normal levels of alanine and aspartate aminotransferase. Erythrocyte sedimentation rate was slightly raised at 35 mm/h. Protein electrophoresis showed a slight increase in alpha-1-globulin and beta-2-globulin and no light chain proteins were found in urine. Anti-nuclear antibodies (ANA) were above the normal value (640 with a normal range < 160) and the ganglioside screening revealed an elevation in anti-GD1a IgG antibodies (109% for a normal range < 50%). Anti GM1, GM2, GD1b, GQ1b, SGPG, MAG as well as GD1a IgG and IgM were normal.

An almost complete neurological recovery was present one month

later after intense physical therapy.

Discussion

A few days following Listeria meningoencephalitis, this patient presented an acute hetereogeneous polyradiculoneuritis affecting the upper arms and sparing the sensory function. She had, as well, a mild taste modification, which suggested a mild cranial nerve dysfunction (probably chorda tympani) and presented a transient tachycardia suggesting mild dysautonomia. The finding of anti-GD1a IgG antibodies strengthens the hypothesis of an acute motor axonal neuropathy (AMAN) subtype of Guillain-Barre syndrome (GBS) [4]. GBS is a heterogeneous disorder characterized by autoimmune peripheral nerve damage with different subtypes including acute inflammatory demyelinating polyneuropathy (AIDP), acute motor axonal neuropathy (AMAN) and acute motor and sensitive axonal neuropathy (AMSAN) [5]. The AMAN type is more commonly encountered in Asia such as Japan or China than in Europe whereas the AIDP is normally more ubiquitous [5,6]. The pathophysiological mechanisms underlying the immune response are still poorly understood. An infectious origin with a cross-reaction between pathogen and a structural resemblance of a host-tissue known as “molecular mimicry” is the most common assumption implicating both cellular and humoral immune systems [6]. One of the supposed mechanisms of nerve damage is a direct dysimmune attack on gangliosides – sialic acid-containing glycosphingolipids – ubiquitously found in tissues and body fluids, but most abundant in the nervous system [4,7]. On the cell surface, gangliosides are involved in cell–cell recognition, adhesion and signal transduction. Anti-ganglioside antibodies and more specifically anti-GD1a and anti-GM1 IgG antibodies have been strongly associated with the axonal component of the neuropathy [4,8]. As described by Ho et al., anti-GD1a IgG are found in 60% of AMAN [4]. In general, the commonest infections associated with GBS are upper respiratory infections or gastroenteritis.

Viruses such as Epstein-Barr virus, cytomegalovirus or hepatitis have also been described [5,8]. In this case, positive anti-HEV IgM antibodies were detected suggesting a recent infection. However, in all cases of GBS related to a hepatitis E infection, ALT and AST levels were at least 1.5 times above the upper limit which is not the case of this patient [9,10]. The differential diagnosis of neuralgic amyotrophy associated with HEV (Parsonage-Turner syndrome) was not considered regarding the absence of pain and amyotrophy. New pathogens presumably involved in the pathogenesis of sporadic cases of GBS are constantly being described especially during outbreaks or epidemics, as recently suggested for Zika virus [11]. Since no data was found on GBS and previous *Listeria monocytogenes* infection, one could suggest a similar molecular mimicry mechanism from a GD1a epitope in the Listeria microorganism triggering the immune response and hence the neurological deficit rapidly recovered after the appropriate antibiotic therapy. GBS can lead to an inability to walk and even to death if untreated. The prognosis for AMAN subtype is much better with rapid improvement of muscle strength [5].

Electrophysiological studies are essential to confirm the diagnosis and to define the GBS subtype. In the classical form (AIDP), the earliest findings are generally prolonged F-wave latencies or poor F-wave repeatability due to demyelination of the nerve roots, followed by temporal dispersion or conduction block [12]. Electrodiagnostic features of AMAN are significantly reduced distal amplitudes in the first few days and partial conduction blocks [5]. In the AMSAN subtype, the sensory potentials are often reduced in amplitude or absent [5,12].

Conclusion

This patient presented meningoencephalitis due to *Listeria monocytogenes* followed by an AMAN subtype of GBS 72 h after the initial symptoms. A dysimmune process leading to axonal dysfunction based on the concept of “molecular mimicry” is suspected. To our knowledge no previous peripheral radiculoneuropathy linked to *Listeria monocytogenes* has ever been reported. An exhaustive screening found no other explanation for this peripheral neuropathy. New pathogens responsible for concomitant or preceding infection associated with sporadic cases of GBS are regularly reported. One should pay special attention for neurologic deficits after infectious diseases in the first 4 weeks after the symptom onset and anti-ganglioside antibodies can be screened in a suggestive clinical situation despite the imperfect specificity and sensitivity. Electrophysiological studies are necessary for accurate diagnosis, subtype classification and establishing prognosis. Immune treatments should be guided by the severity of neurological deficits and are often not needed in the AMAN subtype.

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Competing interests

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Authors’ contribution

All contributing authors were involved in the care of the patient. Nathalie Tebib drafted the initial paper. All authors participated in literature research and extension of the initial draft. All authors agreed on the final submitted version.

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