

Isolated musculocutaneous involvement in neuralgic amyotrophy associated with SARS-CoV2 vaccination

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Dear Editor,

Neuralgic amyotrophy (NA) or Parsonage-Turner syndrome is a clinical syndrome typically characterized by sudden pain attacks in the shoulder and upper arm, followed by patchy muscle paresis in the upper limb. The isolated musculocutaneous nerve involvement is extremely rare [1]. Neuralgic amyotrophy is commonly preceded by a trigger event, such as infection, surgery, and, in some rare cases, vaccination [1]. Although the severe acute respiratory syndrome coronavirus-2 (SARS-COV-2) vaccine has been proven to be safe in the early registration phase, recently significant neurological side effects have been referred [2]. We describe a case of NA with isolated musculocutaneous nerve involvement which developed a few days after administration of this vaccination.

A 50-year-old healthy female on 1st June 2021 received her first dose of the SARS-CoV-2 BNT162b2 vaccination. Two days later, she developed sudden onset severe pain in the left shoulder. The day after, the pain moved to the right shoulder then disappeared. On 11th June 2021, when she was on holiday in Tuscany, she complained again of severe pain in her left arm, more pronounced on the shoulder and elbow. For this reason, she was admitted to the emergency department at "Azienda Usl Toscana Nord Ovest – Ospedale di Cecina." Neurological examination was unremarkable with the exception of weakness (MRC Medical Research Council 3/5) in the right biceps brachii. The sensation was diminished to light touch throughout the right C5 dermatome.

³ Neurological Rehabilitiation, Policlinico San Marco, Gruppo San Donato, Zingonia, Bergamo, Italy Left biceps brachii tendon reflex was decreased. Blood tests were unremarkable, in particular: Herpes simplex 1-2 and Varicella zoster viral immunoglobulin M titers, C-reactive protein, creatine kinase levels, liver enzymes, serum and urine electrophoresis with immunofixation, serum-free light chains, HIV antibody, Lyme antibody, sedimentation rate, rheumatoid factor, antinuclear antibody, extractable nuclear antigen panel/screen, antineutrophil cytoplasmic antibodies, complement levels, and serum paraneoplastic antibody profile. MRI of the cervical spine was normal. On 23rd June 2021, the first electromyography and nerve conduction studies (NCS) were performed. The motor NCS were in the normal range (Supplementary Table S1) while there was a decrease in the recruitment pattern in the left biceps brachii at the needle examinations. On 24th July 2021, because of the persistence of pain, a second neurophysiological evaluation was executed: Nerve conduction studies showed the absence of the sensory nerve action potential (SNAP) (Fig. 1) for the left lateral antebrachial cutaneous sensory nerve and the compound motor action potential (CMAP) at the biceps brachii was absent (Supplementary Table S1) (Fig. 1). Furthermore, 3 + fibrillations and 3 + positive sharp waves were present at the right biceps brachii as well as the absence of the voluntary recruitment pattern. Treatment with oral prednisone 25 mg/d was immediately started. Pregabalin up to 400 mg/d was also started with actual significant improvement in pain and slight improvement of strength. Subsequently, steroids were tapered to 5 mg/d over 5 weeks and occupational therapy commenced to maintain range of motion and facilitate ADLs.

We describe a case of NA with isolated musculocutaneous nerve involvement following administration of the SARS-CoV-2 BNT162b2 vaccine. Although there have been seven reports of brachial plexopathies [see, for example, references 3 and 4], to the best of our knowledge, this represents the first description of isolated musculocutaneous nerve involvement. Our patient's clinical presentation, electrodiagnostic findings of focal involvement of the

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Fig. 1 On 23rd June 2021 (T0), nerve conduction studies (NCS) were performed. The motor and sensory NCS were in the normal range. On 24th July 2021 (T1), NCS showed the absence of the compound

musculocutaneous nerve with the absence of the lateral antebrachial cutaneous sensory nerve SNAP (representing the gold standard in musculocutaneous lesions) alongside the partial improvement of symptoms after initiation of steroids, supports the diagnosis of NA with isolated musculocutaneous nerve involvement. Neuralgic amyotrophy represents a rare disorder; its overall incidence is nowadays estimated to be about 1 in 1000 [1], but isolated musculocutaneous involvement is anecdotal. Post-vaccination NA is also very rare, as a search in the VAERS (Vaccine Adverse Event Reporting Database) brought to light. The database revealed from 2018 to 2020 amidst 350 million seasonal influenza vaccinations that 18 cases of brachial neuritis [5] (classified as "brachial plexopathy," "brachial plexus injury," or "brachial radiculitis") were found. This database in a similar number of SARS-CoV-2 vaccinations (358.599.835 doses with at least one dose in 200.000.000) reported 85 cases of brachial neuritis [5] (classified as "neuralgic amyotrophy" 64 patients, "brachial plexopathy" 16 patients, "brachial plexus injury" 5 patients). Until now, few cases of brachial plexopathy have been published [see,

motor action potential at the biceps brachii and of the sensory nerve action potential for the left lateral antebrachial cutaneous sensory nerve

for example, references 3 and 4]. Obviously, our report has no epidemiological meaning as up to now more than 4.8 billion SARS-CoV-2 vaccine doses have been administered globally; thus, it remains to be seen whether the incidence of SARS-CoV-2 BNT162b2 vaccination triggered NA or other rare neurological side effects more than was expected as some authors point out [2]. In any case, further studies are needed to determine whether a causal relationship exists between these vaccines and neurological sequelae. Vaccination is currently the most effective strategy for fighting COVID; understanding and defining its side effects will facilitate safety assessments and minimize vaccination hesitation among the general population.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s10072-022-06004-z.

Declarations

Ethical approval and Informed consent Informed consent statement has been obtained from patient.

Conflict of interest The authors declare no competing interests.

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