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LETTER TO THE EDITOR

# A rare case of pure sensitive Parsonage-Turner syndrome

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Dear Editor,

Parsonage-Turner syndrome (PTS) or neuralgic amiotrophy (NA) is a rare disorder characterised by severe neuropathic pain and subsequent patchy paresis in the upper extremity. It is usually monolateral, but bilateral involvement is reported in up to 15% of patients. There are idiopathic and familiar forms (about 200 families known worldwide). It is thought to have an immune-mediated aetiology and several antecedent events that may serve as triggers have been reported [1]. Corticosteroids for 2 weeks seem to shorten the painful phase and provide better recovery after 1 year [2]. The disorder is usually monophasic, but recurrence is reported from 5% to 26%, particularly in familiar forms. Although NA is commonly considered a motor disorder, sensory deficits are reported in most patients, but are usually mild [1].

We reported a rare case of bilateral pure sensitive neuralgic amiotrophy developed 2 weeks after TBE vaccination. A 62-year-old woman presented at our electromyography laboratory complaining for 40 days hypoesthesia and paresthesias in bilateral median nerve territory. This disorder followed an episode of severe acute pain localised in bilateral arms and forearms, spontaneously resolved in 2 days. She did not complain any motor deficit or pain localised in cervical region. Her medical history was unremarkable: in particular, she did not report infectious disease, fever and traumas, but she received a TBE-vaccination 2 weeks before the onset of pain. Contrast-enhanced cervical MRI did not reveal significant alterations. Nerve conduction study revealed normal upper limbs CMAPs (compound muscular action potentials), absent SNAPs (sensory nerve action potentials) of bilateral median nerve for I finger and reduced SNAPs amplitudes with normal latency and conduction velocity of right median nerve for II–III fingers and right medial antebrachial cutaneous nerve (MAC). Needle electromyography revealed normal MUPs (motor unit potentials) with normal interferential pattern of recruitment. Data of nerve conductions study are reported in Tables 1, 2, and 3 and for needle electromyography in Table 4. Laboratory tests including CBC, PT, aPTT, INR, RCP, ESR, vitamin B12, folate, glycemia, ANA, anti DNA, RF, anti-neuronal antibodies and neoplastic marker were all normal. According to clinical and neurophysiological data, we concluded for a case of unusual Parsonage-Turner syndrome. No pharmacological treatment was prescribed because the symptom was spontaneously improving and she did not complain of pain. Six months later, SNAPs of bilateral median nerve for I finger were detectable with reduced amplitudes and SNAPs of right median nerve for II–III fingers and MAC were normal. The patient did not complain any symptom.

## Discussion

NA has always been considered a rare disorder (annual incidence of 2/100.000 people), although a recent cohort study revealed an annual incidence of 1/1000 people in primary care [3], suggesting a possible underestimation and poorly knowledge to general practitioners. It is frequently misdiagnosed as compressive cervical root disorder, but also as rotator cuff syndrome and scapular girdle

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**Table 1** Motor nerves conduction study

| | Lat ms | Amp pp mV | Dur tot ms | Area ms*mV | CV m/s | dist mm | Diff amp pp % | Diff area neg % |
|---|---|---|---|---|---|---|---|---|
| Median motor left | | | | | | | | |
| Wrist-APB | 4.02 | 7.1 | 18.7 | 12.3 | | 70.0 | | |
| Elbow-wrist | 7.67 | 7.0 | 18.3 | 12.4 | 57.5 | 210 | −1.41 | −0.81 |
| Median motor right | | | | | | | | |
| Wrist-APB | 3.71 | 8.7 | 16.7 | 14.0 | | 70.0 | | |
| Elbow-wrist | 7.67 | 8.7 | 19.2 | 13.9 | 53.0 | 210 | 0 | −0.71 |
| Ulnar motor right | | | | | | | | |
| Wrist-ADM | 2.25 | 11.8 | 17.9 | 17.9 | | 70.0 | | |
| Bl. elbow-wrist | 5.56 | 11.7 | 20.0 | 17.8 | 65.0 | 215 | −15.0 | −13.9 |
| Ab. elbow-Bl. elbow | 7.75 | 11.4 | 21.9 | 18.0 | 59.4 | 130 | −9.8 | −13.4 |
| Ulnar motor left | | | | | | | | |
| Wrist-ADM | 1.88 | 10.9 | 15.5 | 18.3 | | 70.0 | | |
| Bl. elbow-wrist | 5.60 | 11.4 | 19.1 | 18.1 | 56.5 | 210 | 4.6 | −1.09 |
| Ab. elbow-Bl. elbow | 7.83 | 11.3 | 22.0 | 18.0 | 53.8 | 120 | −0.88 | −0.55 |

myopathy. Some evidences suggest the usefulness of corticosteroid treatment [2] during the first painful phase, but treatment is often limited by too late diagnosis of PTS. Also for a neurologist diagnosing PTS may not be immediate: it is easy for the typical form with monolateral shoulder pain followed by atrophy of upper limb muscles, but atypical forms could be misinterpreted as brachial infiltrative plexopathy, vasculitis and also for cervical radiculopathy. We reported a case with some rare characteristics: bilateral and parcellar sensory involvement. Principal differential diagnoses included:

– cervical root compression: this hypothesis was excluded for numerous reason: bilateral presentation, normal cervical MRI and post-ganglionic involvement evidenced by neurophysiology;
– infiltrative plexopathy: we excluded this hypothesis for the absence of history of malignancies, bilateralism of symptoms and minimal involvement (only median nerves and right MAC);

**Table 2** F waves study

| F wave | | | |
|---|---|---|---|
| | M lat ms | lat f min ms | f-m lat ms |
| Median left | | | |
| Wrist-APB | 3.8 | 27.1 | 23.3 |
| Ulnar left | | | |
| Wrist-ADM | 2.1 | 26.0 | 23.9 |

**Table 3** Sensory nerves conduction study

| | Lat ms | cv m/s | Distance mm | Amp uV |
|---|---|---|---|---|
| Sensitives-right | | | | |
| Wrist-I fing Rad | 1.45 | 69.0 | 100 | 7.0 |
| Wrist-I fing Med | – | – | – | – |
| Wrist-II fing Med | 2.06 | 68.0 | 140 | 12.5 |
| Wrist-III fing Med | 2.58 | 58.1 | 150 | 17.4 |
| Wrist-IV fing Med | 2.35 | 59.6 | 140 | 13.4 |
| Wrist-IV fing Uln | 2.23 | 62.8 | 140 | 11.2 |
| Wrist-V fing Uln | 1.91 | 62.8 | 120 | 13.2 |
| Lateral antebrachial cutaneous | 1.51 | 69.5 | 105 | 10.9 |
| Medial antebrachial cutaneous | 1.55 | 71.0 | 110 | 4.7 |
| Sensitives-left | | | | |
| Wrist-I fing Rad | 2.03 | 56.7 | 115 | 7.3 |
| Wrist-I fing Med | – | – | – | – |
| Wrist-II fing Med | 2.97 | 48.8 | 145 | 3.6 |
| Wrist-III fing Med | 3.10 | 50.0 | 155 | 2.0 |
| Wrist-IV fing Med | 2.55 | 50.9 | 130 | 13.0 |
| Wrist-IV fing Uln | 2,28 | 57,02 | 130 | 13.5 |
| Wrist-V fing Uln | 2.08 | 52.9 | 110 | 15.5 |
| Lateral antebrachial cutaneous | 1.72 | 72.7 | 125 | 13.7 |
| Medial antebrachial cutaneous | 1.56 | 70.5 | 110 | 11.7 |

**Table 4** Needle electromyography study

| Muscle | Spontaneous activity | | Voluntary activity | | | |
|---|---|---|---|---|---|---|
| | Fibrillations | PSW | Amp | Dur | Poly | Recruit |
| Left Abd pollicis brevis | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Right Abd pollicis brevis | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Left biceps | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Right brachioradialis | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Left deltoideus post | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Left ext dig communis | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Left flex dig superfic | 0/10 | 0/10 | Normal | Normal | Normal | Normal |
| Right inteross dors I | 0/10 | 0/10 | Normal | Normal | Normal | Normal |

– brachial plexus vasculitis: it was less probable than NA because all laboratory tests were negative and clinical presentation was atypical (spontaneous resolution of pain, parcelling involvement).

We did not prescribe a plexus MRI because we considered an infiltration improbable and moreover the symptom was spontaneously improving.

NA is considered atypical when is hereditary, non-painful, distal (motor or sensory symptoms distal to the elbow joint), extensive (involving many roots, division cords or nerves of both brachial plexus), extra-brachial, recurrent, purely sensory or autonomic [4]. Sensory complaints are presented in 66% to 78% of cases [1] as part of the classic pattern, but isolated sensory manifestations are very rarely reported. The largest case series describes 8 patients with monolateral involvement of LAC and/or median nerve [5].

## Conclusion

We describe this rare presentation of PTS to underline the importance of consider this disorder also in cases of atypical presentation, in order to recognise and treat it promptly, avoiding unnecessaries exams or treatments. At our knowledge, this is the first case reported of PTS developed after a TBE vaccination.

## Compliance with ethical standards

**Conflict of interest** The authors declare that they have no conflict of interest.

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