

Parsonage Turner syndrome of the brachial plexus secondary to Covid-19 vaccine: A case report

Islam Mejri¹, Lenda Ben Hmida², Ines Bedoui², Tasnim Znegui², Maroua Kacem ², Zied Moatemri², Hajer Derbali², Samia Jammeli ², and Nejiba Fekih Mrissa ²

¹University of Tunis El Manar Faculty of Sciences of Tunis

²University of Tunis El Manar

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Abstract

Parsonage Turner syndrome (PTS) is a peripheral inflammatory neuropathy of unknown etiology. We present a rare case of a patient with PTS post-covid-19 BNT162b2 mRNA vaccine. Symptoms occurred fifteen days after the second dose. The patient was treated with corticosteroids, analgesics and physical rehabilitation with a partial recovery.

Title page

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Mejri Islam^{1,3}, Lenda Ben Hmida^{1,3}, Ines Bedoui^{2,3,4}, Tasnim Znegui^{1,3}, Kacem Maroua¹, Zied Moatemri^{1,3}, Hager Derbali^{2,3}, Samia Jammeli ^{3,5}, Fekih Mrissa Nejiba ^{4,3}

1 Pneumology Department, Military Hospital of Tunis, Tunisia

2 Neurology Department, Military Hospital of Tunis, Tunisia

3 Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

4 Molecular Biology Unit (UR17DN06), Laboratory of hematology, Military Hospital Of Tunis , Tunisia

5 Department of rheumatology, Rabta Hospital, Tunisia

Mejri Islam: Doctor, islammejri@gmail.com, Pneumology Department, Military Hospital of Tunis, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia.

Lenda Ben Hmida: Doctor, benhmidalenda@gmail.com, Pneumology Department, Military Hospital of Tunis, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Ines Bedoui : Doctor, bedoui.ines@hotmail.fr, Molecular Biology Unit (UR17DN06), Laboratory of hematology, Military Hospital Of Tunis , Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Tasnim Znegui : Doctor, Zneguitasnim@gmail.com, Pneumology Department, Military Hospital of Tunis, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Kacem Maroua : Doctor, mriwa001@gmail.com, Pneumology Department, Military Hospital of Tunis, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Zied Moatemri: Professor Doctor, zied.moatemri@fnt.utm.tn, Pneumology Department, Military Hospital of Tunis, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Hager Derbali : Doctor, hajer.derbali@hotmail.fr, Neurology Department, Military Hospital of Tunis, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Samia Jammeli : Doctor, samia.jemmali@fmt.utm.tn, Department of rheumatology, Rabta Hospital, Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Fekih Mrissa Nejiba : Professor Doctor, fnajiba@yahoo.fr, Molecular Biology Unit (UR17DN06), Laboratory of hematology, Military Hospital Of Tunis , Faculty of medicine of Tunis, University Tunis El Manar, Tunisia

Key Clinical Message

We highlight the immunological hypothesis of PTS post-covid-19 BNT162b2 mRNA vaccine.

The prevalence of PTS post-vaccination is most likely to increase with covid-19 vaccines.

Early recognition is necessary for adequate treatment.

Abstract

Parsonage Turner syndrome (PTS) is a peripheral inflammatory neuropathy of unknown etiology. We present a rare case of a patient with PTS post-covid-19 BNT162b2 mRNA vaccine. Symptoms occurred fifteen days after the second dose. The patient was treated with corticosteroids, analgesics and physical rehabilitation with a partial recovery.

Key Words: Parsonage Turner syndrome, COVID-19, vaccine

Background

Parsonage Turner syndrome (PTS), also known as neuralgic amyotrophy or idiopathic brachial plexopathy, is a rare peripheral multifocal inflammatory neuropathy that usually affects the upper limbs(1). However, it is widely misdiagnosed because of its heterogeneous clinical appearance(2). The classic presentation is a patient with subacute onset of asymmetric shoulder pain, followed several days later by weakness and amyotrophy(3). Although its exact cause is still unknown, multiple factors have been identified such as immunological (infection, vaccination, surgery, pregnancy, physical or mental stress), mechanical (trauma, sports or heavy labor) and genetic factors(4). In the last years PTS has been associated with severe covid-19 infection as well as its vaccination(5). Therefore, we report a rare case of a sub acute onset of PTS as a result of Covid-19 vaccine.

Case description

We represent the case of a 50-year-old right-handed male patient, admitted to the neurology department of the military hospital of Tunis-Tunisia. He has no medical history of chronic diseases or medication use. He had a mild covid-19 infection confirmed by RT-PCR on April 2021. He had a complete immunization schedule with no history of vaccine reaction. He received the first dose of Covid-19 BNT162b2 mRNA vaccine on the 8th August 2021 followed by the second dose on the 4th December 2021, both in his right deltoid muscle. There was no recent trauma, surgery or infectious disease. Fifteen days after the second dose of the vaccine, he presented with pain on the injection site, resistant to pain relievers, radiating to the right shoulder and the neck. Two weeks later, he presented numbness, heaviness and muscle weakness of the upper right limb. There were no sensory disturbances or other symptoms. General physical examination was normal. Neurological examination revealed hypoaesthesia and monoparesis of the right upper limb with muscle weakness in shoulder abduction and extension and right deltoid amyotrophy. No motor deficits were found in other parts of the body. All deep tendon reflexes were normoactive and symmetrical. Superficial and deep sensation was normal. His laboratory results were all normal, as well as the lumbar puncture results. Electroneuromyography (ENMG) performed 30 days from symptoms' onset showed signs of brachial plexus neuritis (neurogenic tracing of the right deltoid muscle). Computed tomography (CT)-scan imaging of the brain was normal. Magnetic resonance imaging (MRI) of the right shoulder, performed two weeks after symptoms' onset, revealed no abnormal results.

Considering all these findings the patient was diagnosed with subacute PTS post-Covid-19 BNT162b2mRNA vaccine. He was treated with corticosteroids grade 2 and analgesics with initiation of physical rehabilitation. During the follow-up period, the patient's medical condition improved; with a partial recovery in motor functions.

Discussion

PTS is the classic presentation of the brachial plexus inflammation, involving the long thoracic, subscapular, superficial radial and anterior interosseous nerves, but can also involve other peripheral nerves, lower brachial plexus and phrenic nerves(1)(6).

PTS is a not a rare neurological disorder as previously thought(1). A recent prospective study suggests that its actual incidence rate is 1 per 1000 per year(7). It is most likely underdiagnosed due to its misleading manifestations (6).

In general, the idiopathic form of PTS occurs mainly in men with a sex ratio of 2 and a median age of 40 years. It is typically characterized by the onset of sudden severe shoulder pain, developing paresis and tingling within several hours to days later, as reported in our case(6).

The diagnosis of PTS is primarily clinical, there is no specific diagnostic test (1). ENMG may lack sensitivity and accuracy in confirming the diagnosis(6). In our case, the pattern brachial plexus neuritis comforted the diagnosis hypothesis. In fact, this sign is only found in 30 to 45% of confirmed PTS cases(4). Shoulder MRI helpsexcluding differential diagnoses such as intrinsic shoulder disorder. Our patient showed no signal abnormalities.

The exact mechanism in PTS is still unknown but multiple factors are involved. The immunological hypothesis is plausible by the fact that 50% of PTS patients have a history of a trigger event such as infection, vaccination, surgery, pregnancy, physical or mental stress(4). Concomitant hepatitis E viral infection was found in 10% of patients which pleads in favor of an infectious or post-infectious mechanism(4). In peripheral nerve biopsy, epineural perivascular mononuclear T-cell infiltration was found(6) which supports the immunological theory.

In this regard, our case is interesting as the symptoms' onset occurred after a vaccination: Covid-19 BNT162b2 mRNA vaccine (manufactured by Pfizer).

Post vaccination PTS is very rare(8). PTS onset occurs within 28 days after vaccination, in an estimated 4.3–15.5% of cases(9). Our patient typically developed classic symptoms of PTS 15 days after receiving the second dose of BNT162b2 mRNA vaccine, as it was reported in the literature.

Concerning covid-19 vaccines, and after at least one dose, 56 reports of PTS were detected by The Vaccine Adverse Event Reporting System (VAERS) in July 2021, among which 24 reports concerned BNT162b2 mRNA vaccine(9).

To date, there is no specific treatment for PTS. Support therapy including corticosteroids, analgesics, immobilization and physical therapy are the milestone of PTS treatment, as it was highlighted in our case(4). No recommendations have been established due to the lack of Randomized Controlled Trials. Intravenous corticosteroids and immunoglobulins can be used in severe extensive PTS with intense pain in order to minimize symptoms duration and recover functional abilities(4).

Outcomes are heterogenous and depend on the phase in which the patient is diagnosed, the intensity of pain, the extent of plexus involvement and whether the symptoms are bilateral or unilateral(10).

Conclusion

PTS is not a rare disease as it was previously considered. It's rather misdiagnosed due to its variable symptoms. An immune system trigger is usually found. The Covid-19 BNT162b2 mRNA vaccine, like other vaccines, can be associated to PTS with a typical presentation. As COVID-19 vaccination rates increase,

it is quite possible that post-vaccination PTS will increase. Thus, early recognition is essential to initiate adequate treatment, leading to better recovery without sequelae.

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Competing interests

The authors declare no competing interest.

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Authors' contributions

All authors have contributed to this work. All authors have read and agreed to the final manuscript.

References

1. IJspeert J, Janssen RMJ, van Alfen N. Neuralgic amyotrophy. *Curr Opin Neurol*. 2021 Oct 1;34(5):605–12.
2. Kim SI, Seok HY, Yi J, Cho JH. Leg paralysis after AstraZeneca COVID-19 vaccination diagnosed as neuralgic amyotrophy of the lumbosacral plexus: a case report. *J Int Med Res*. 2021 Nov;49(11):3000605211056783.
3. Crespo Burillo JA, Lorienté Martínez C, García Arguedas C, Mora Pueyo FJ. Amyotrophic neuralgia secondary to Vaxzevri (AstraZeneca) COVID-19 vaccine. *Neurol Barc Spain*. 2021 Sep;36(7):571–2.
4. Kim TU, Chang MC. Neuralgic amyotrophy: an underrecognized entity. *J Int Med Res*. 2021 Apr;49(4):3000605211006542.
5. Vitturi BK, Grandis M, Beltramini S, Orsi A, Schenone A, Icardi G, et al. Parsonage-Turner syndrome following coronavirus disease 2019 immunization with ChAdOx1-S vaccine: a case report and review of the literature. *J Med Case Reports*. 2021 Dec 13;15(1):589.
6. Van Eijk JJJ, Groothuis JT, Van Alfen N. Neuralgic amyotrophy: An update on diagnosis, pathophysiology, and treatment. *Muscle Nerve*. 2016 Mar;53(3):337–50.
7. van Alfen N, van Eijk JJJ, Ennik T, Flynn SO, Nobacht IEG, Groothuis JT, et al. Incidence of neuralgic amyotrophy (Parsonage Turner syndrome) in a primary care setting—a prospective cohort study. *PloS One*. 2015;10(5):e0128361.
8. Mahajan S, Zhang F, Mahajan A, Zimnowodzki S. Parsonage Turner syndrome after COVID-19 vaccination. *Muscle Nerve*. 2021 Jul;64(1):E3–4.
9. Queler SC, Towbin AJ, Milani C, Whang J, Sneag DB. Parsonage-Turner Syndrome Following COVID-19 Vaccination: MR Neurography. *Radiology*. 2022 Jan;302(1):84–7.
10. Alvarado M, Lin-Miao Y, Carrillo-Arolas M. Parsonage-Turner syndrome post-infection by SARS-CoV-2: a case report. *Neurol Barc Spain*. 2021 Sep;36(7):568–71.

Abbreviation:

PTS: Parsonage-Turner Syndrome

ENMG: Electroneuromyography

MRI: Magnetic resonance imaging