Since January 2020 Elsevier has created a COVID-19 resource centre with free information in English and Mandarin on the novel coronavirus COVID-19. The COVID-19 resource centre is hosted on Elsevier Connect, the company's public news and information website.

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Parsonage-Turner syndrome after COVID-19 infection

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As our understanding of severe acute respiratory syndrome coronavirus (COVID-19) infections continues to evolve, it has become apparent that the clinical manifestations of the disease are not strictly limited to the respiratory system. In rare cases, severe neurological sequelae have followed viral infection. When these symptoms affect the brachial plexus, significant dysfunction of the shoulder and upper extremity can result.[7]

Parsonage-Turner syndrome, also known as neuralgic amyotrophy, is a rare, immune-mediated inflammatory process that is characterized by the acute onset of upper extremity pain proceeded by progressive neurological deficits including weakness and atrophy.[4,9,10] The most frequently affected portions of the brachial plexus include the suprascapular, musculocutaneous, radial, long thoracic, and axillary nerves. Manifestations of weakness vary but typically include difficulty with flexion, external rotation, and internal rotation. Rare cases may result in scapular winging. Pain may be made worse with activity and can often result in sleep disturbances.[10]

The exact cause of Parsonage-Turner syndrome is unknown. The most widely acknowledged causes are postinfectious and autoimmune sequelae.[9] It is also thought that there may be a genetic component as well, as one in every ten patients with Parsonage-Turner syndrome reports a family history of the condition.[3] Because of the complexity of the condition, neuralgic amyotrophy is a clinical diagnosis, often diagnosed only after ruling out several possible conditions with similar presentations, as no one test exists that can either confirm or deny the diagnosis with absolute certainty. Overuse of the trapezius, pectoralis muscles, and levator scapulae is common in patients with neuralgic amyotrophy to compensate for weakness elsewhere, and this may result in associated hypertrophy of the aforementioned musculature.[17] Advanced imaging and diagnostic tests are frequently used in conjunction with a high index of clinical suspicion as a diagnosis for neuralgic amyotrophy. These tests include electromyography (EMG) and magnetic resonance imaging (MRI). The commonly discussed MRI findings in the setting of Parsonage-Turner include subacromial bursitis, edema within the rotator cuff musculature, and atrophy of the affected muscles, although this may be a late finding.[17]

Few case reports exist of documented Parsonage-Turner syndrome subsequent to a COVID-19 infection,[6] and the authors found no case reports of Parsonage-Turner syndrome subsequent to a COVID-19 infection described to date in the orthopedic literature. As such, the authors report a case of Parsonage-Turner syndrome after a COVID-19 infection resulting in profound weakness because of neuropathy of the upper trunk of the brachial plexus.

Case details

This is a case of a 61-year-old man with an acute onset of pain and weakness affecting his left shoulder after a COVID-19 infection. Before his initial presentation in the orthopedic clinic, the patient was admitted to the hospital with hemoptysis after two days of a worsening cough. His symptom onset corresponded to a time of increased COVID-19 incidence within the region. The patient had undergone a pulmonary computed tomography scan on arrival to the emergency department (ED). This was positive for a small, acute pulmonary embolism and a small infiltrate in the right upper lobe with associated lymphadenopathy. Laboratory values at that time were largely normal. Given his symptoms, the patient was tested for COVID-19 and found to be positive. At the time of admission, he did not meet the criteria for treatment with convalescent plasma as his respiratory symptoms were relatively mild. As such, his initial treatment was primarily supportive. Given his comorbidities (hypertension, prior arrhythmias), the patient was started on

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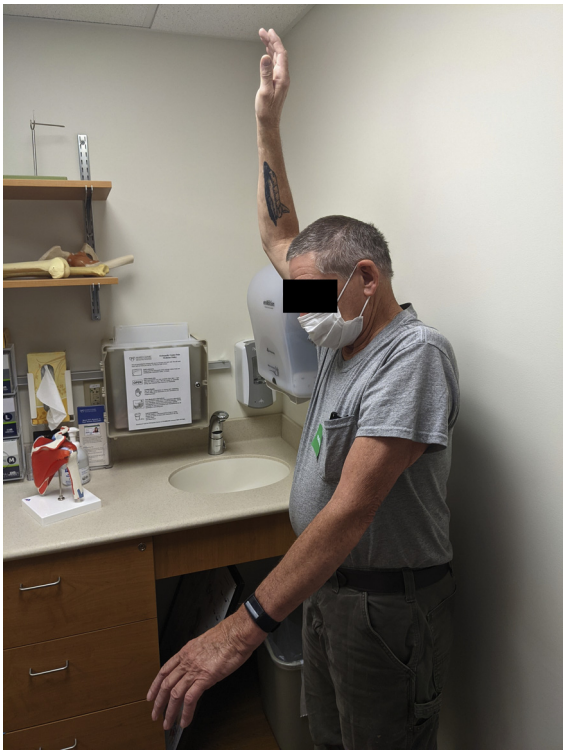

Figure 1 Profound weakness with attempted active forward elevation of the affected, *Left* arm.

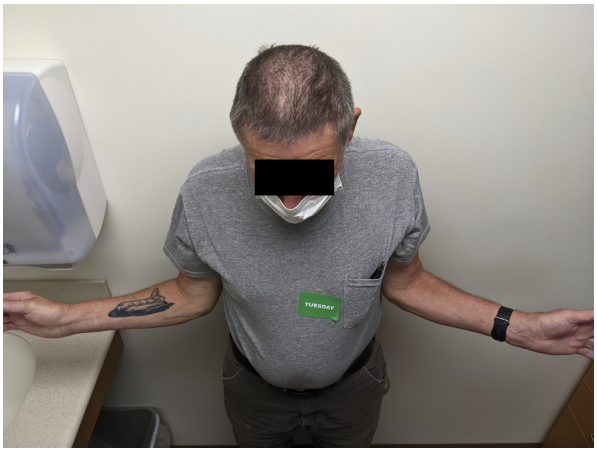

Figure 2 Preserved active external rotation when no resistance is applied.

Figure 3 Preserved subscapularis function with belly press testing with and without resistance.

remdesivir and was subsequently discharged 3 days after admission in stable condition after gradual improvement in his respiratory status. Patient consent was obtained for this publication.

At his initial follow-up with his primary care physician (PCP), he was noted to have resolution of his respiratory complaints. At his subsequent follow-up, he complained of leg weakness and back pain—including a day's history of bilateral leg weakness/anterior thigh weakness. He had associated burning-type pain in the lower extremity which was quite severe and limited his ability to sleep. He denied any trauma, fall, or other clear causes of his lower extremity and back complaints. Although the pain was initially quite severe, it gradually resolved over the course of several weeks.

Several weeks later, the patient presented to the emergency room with the insidious onset of severe shoulder pain. At that time, he stated that his shoulder pain had been getting progressively worse over a period of hours, again describing a burning-type pain, limiting his sleep. Ibuprofen, muscle relaxers, and narcotic pain medications did little to alleviate his symptoms at that time. The patient described the pain as excruciating and localized it to the scapular region. In this instance, the pain was not alleviated or aggravated by shoulder movement. He was directed by the ED to follow-up with his primary care physician for further evaluation. At a one-week follow-up visit with primary care, the patient was still reporting significant shoulder pain and was referred to orthopedics for further evaluation, as he had noted progressive weakness and loss of active motion.

At the time of initial presentation to orthopedics, the patient's pain had largely resolved, but his profound weakness and related shoulder dysfunction had persisted and worsened. The patient had significant limitations of active range of motion with a physical examination demonstrating profound weakness with forward flexion and significant weakness with external rotation. He did have preserved subscapularis function against resistance and a full passive range of motion in all planes (Figs. 1-3). He had sensory changes at that time with side-to-side differences in sensation in the area of the axillary nerve distribution, overlying the lateral shoulder. X-rays obtained at that time were largely benign, demonstrating mild glenohumeral arthritis without significant elevation of the humeral head.

The patient had a history of a prior rotator cuff repair performed 2.5 years previously, with an uneventful recovery resulting in normal strength and range of motion at 6 months from surgery, but given his prior procedure, an MRI was performed to rule out a recurrent tear—although his presentation would have been atypical given the severity of his pain and its insidious onset without a clear traumatic cause. The MRI demonstrated a well-healed rotator cuff. The only significant findings at that time were mild edema and atrophy of the supraspinatus and deltoid. At that point, the primary concern shifted to a neurologic etiology for his profound weakness (Fig. 4).

As such, an EMG was ordered. This demonstrated evidence of a left brachial plexopathy primarily affecting the upper trunk with evidence of ongoing denervation. There was a severe reduction in recruitment in the infraspinatus, deltoid, and, to a lesser extent, the biceps brachii with associated fibrillations. He had a cervical spine MRI performed previously, which was benign. Physical therapy was initiated with a concomitant home exercise regimen. His pain remained relatively mild both at rest and with activity. Unfortunately, his weakness improved only very gradually, and at 8 months

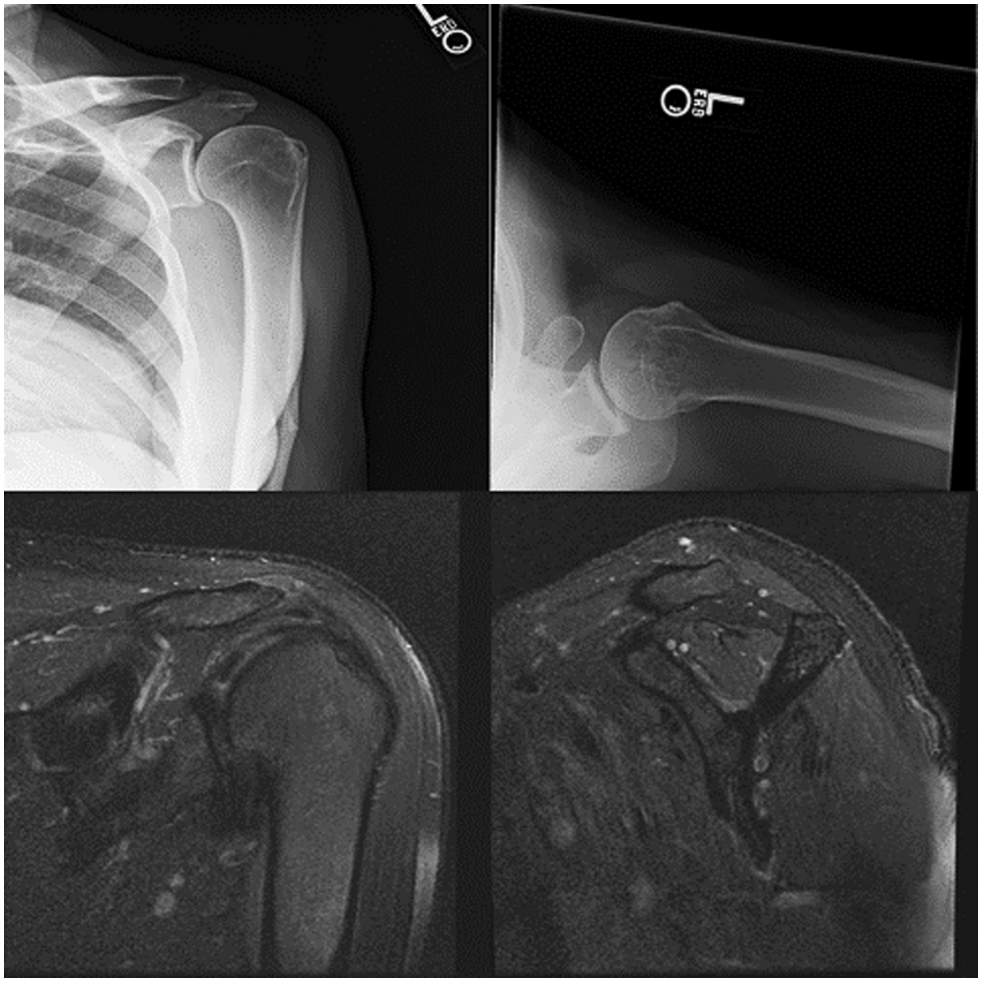

Figure 4 Grashey and axillary X-rays of the patient demonstrating mild glenohumeral arthritis. The T2-fat–suppressed MRI image demonstrating mild edema within the deltoid and supraspinatus muscle belly. *MRI*, magnetic resonance imaging.

from the time of the symptom onset, he had significant difficulties with activities over shoulder height. Owing to his physically demanding job, this weakness significantly limited his ability to work.

Given his continued limitations at 8 months from the original presentation, the possibility of a tendon transfer was discussed to compensate for his loss of forward elevation and external rotation. This was discussed in the setting of continued lack of improvement at or after 1 year from the symptom onset. At that time, the patient was content to proceed with watchful waiting and continuing with home exercises, with the understanding that the return of function after Parsonage-Turner syndrome can be quite prolonged.[8] At his follow-up visit 1 year after the symptom onset, he had noted continued improvements in function, with persistent overhead weakness, and had returned to working as a plumber. As such, no further intervention was indicated at that time.

Discussion

Parsonage-Turner syndrome is a rare, complex condition with many potential causes. Historically, brachial plexus neuritis has been seen in the postviral setting (among many other scenarios), as it was seen in this case after a confirmed COVID-19 infection. The diagnosis of Parsonage-Turner syndrome is typically a diagnosis of exclusion—established only after ruling out several related causes of upper extremity dysfunction. Treatment commonly involves the care of multiple specialists and frequently involves evaluation with electrodiagnostic studies. If limitations persist, tendon transfer surgery may be considered—with the exact procedure being tailored to the specific pathology. In this instance, one might have considered either a latissimus dorsi or an upper trapezius transfer.

Conservative management with analgesics and physical therapy with a focus on the range of motion and strengthening exercises is generally the only treatment required, although time to recovery can be quite protracted and full recovery is variable.[1,2,12,14] Previous medical treatments have included corticosteroids, although this has only been evaluated in a limited sense and with variable outcomes.[16] The optimal duration of physical therapy is unknown; however, several studies show that recovery often requires around 6 to 12 months[11,13-15,18] with full recovery taking up to 3 years.[8]

In addition to recent incidents of neurologic sequelae after a confirmed case of COVID-19, there are limited reports of neuralgic amyotrophy after COVID-19 vaccination.[5] While the long-term effects of COVID-19 continue to be investigated, it has become apparent that a multitude of associated conditions are possible. These can be seen both after vaccination and subsequent to viral infection.

Conclusion

This was a case of neuralgic amyotrophy associated with a COVID-19 infection. The patient had migrating neuralgia initially,

which culminated in an ED visit for severe shoulder pain—this was recalcitrant to oral and IV pain medications and was associated with weakness and loss of motion. The patient improved slowly over the course of a year, although he had persistent weakness overhead at his last follow-up.

In the setting of Parsonage-Turner syndrome, the use of EMG studies can be helpful in the diagnosis of the condition and can also be valuable in delineating recovery (or lack thereof). MRI studies can be helpful in ruling out related pathology and may demonstrate edema within the muscle bellies of affected musculature. Recovery can be prolonged, and watchful waiting is typically the treatment of choice, with associated physical therapy to maintain motion. If no significant recovery is achieved at 12-18 months from the symptom onset, tendon transfers may represent a viable option for restoring motion.

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References

1. Cup EH, Ijspeert J, Janssen RJ, Bussemaker-Beumer C, Jacobs J, Pieterse AJ, et al. Residual complaints after neuralgic amyotrophy. Arch Phys Med Rehabil 2013;94:67-73. https://doi.org/10.1016/j.apmr.2012.07.014.
2. Gaskin CM, Helms CA. Parsonage-Turner syndrome: MR imaging findings and clinical information of 27 patients. Radiology 2006;240:501-7. https://doi.org/10.1148/radiol.2402050405.
3. Gstoettner C, Mayer JA, Rassam S, Hruby LA, Salminger S, Sturma A, et al. Neuralgic amyotrophy: a paradigm shift in diagnosis and treatment. J Neurol Neurosurg Psychiatry 2020;91:879-88. https://doi.org/10.1136/jnnp-2020-323164.
4. Ismail II, Abdelnabi EA, Al-Hashel JY, Alroughani R, Ahmed SF. Neuralgic amyotrophy associated with COVID-19 infection: a case report and review of the literature. Neurol Sci 2021;42:2161-5. https://doi.org/10.1007/s10072-021-05197-z.
5. Mahajan S, Zhang F, Mahajan A, Zimnowodzki S. Parsonage Turner syndrome after COVID-19 vaccination. Muscle Nerve 2021;64:E3-4. https://doi.org/10.1002/mus.27255.
6. Mitry MA, Collins LK, Kazam JJ, Kaicker S, Kovanlikaya A. Parsonage-turner syndrome associated with SARS-CoV2 (COVID-19) infection. Clin Imaging 2021;72:8-10. https://doi.org/10.1016/j.clinimag.2020.11.017.
7. Pezzini A, Padovani A. Lifting the mask on neurological manifestations of COVID-19. Nat Rev Neurol 2020;16:636-44. https://doi.org/10.1038/s41582-020-0398-3.
8. Seror P. Neuralgic amyotrophy. An update. Joint Bone Spine 2017;84:153-8. https://doi.org/10.1016/j.jbspin.2016.03.005.
9. Smith CC, Bevelaqua AC. Challenging pain syndromes: Parsonage-Turner syndrome. Phys Med Rehabil Clin N Am 2014;25:265-77. https://doi.org/10.1016/j.pmr.2014.01.001.
10. Tjoumakaris FP, Anakwenze OA, Kancherla V, Pulos N. Neuralgic amyotrophy (Parsonage-Turner syndrome). J Am Acad Orthop Surg 2012;20:443-9. https://doi.org/10.5435/JAAOS-20-07-443.
11. Tsairis P, Dyck PJ, Mulder DW. Natural history of brachial plexus neuropathy. Report on 99 patients. Arch Neurol 1972;27:109-17.
12. Turner JW, Parsonage MJ. Neuralgic amyotrophy (paralytic brachial neuritis); with special reference to prognosis. Lancet 1957;273:209-12.
13. van Alfen N. The neuralgic amyotrophy consultation. J Neurol 2007;254:695-704. https://doi.org/10.1007/s00415-006-0246-4.
14. van Alfen N, van der Werf SP, van Engelen BG. Long-term pain, fatigue, and impairment in neuralgic amyotrophy. Arch Phys Med Rehabil 2009;90:435-9. https://doi.org/10.1016/j.apmr.2008.08.216.
15. van Alfen N, van Engelen BG. The clinical spectrum of neuralgic amyotrophy in 246 cases. Brain 2006;129:438-50. https://doi.org/10.1093/brain/awh722.
16. van Alfen N, van Engelen BG, Hughes RA. Treatment for idiopathic and hereditary neuralgic amyotrophy (brachial neuritis). Cochrane Database Syst Rev 2009;2009:CD006976. https://doi.org/10.1002/14651858.CD006976.pub2.
17. Van Eijk JJ, Groothuis JT, Van Alfen N. Neuralgic amyotrophy: An update on diagnosis, pathophysiology, and treatment. Muscle Nerve 2016;53:337-50. https://doi.org/10.1002/mus.25008.
18. Walsworth MK, Mills JT 3rd, Michener LA. Diagnosing suprascapular neuropathy in patients with shoulder dysfunction: a report of 5 cases. Phys Ther 2004;84:359-72.