[Primary Care]

Isolated Unilateral Brachial Neuritis of the Phrenic Nerve (Parsonage-Turner Syndrome) in a Marathon Runner With Exertional Dyspnea

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Parsonage-Turner syndrome, or acute brachial neuritis/plexitis, is a rare condition that should be included in the differential diagnosis of any athlete who presents with antecedent flulike symptoms, with progression to significant neuropathic pain, followed by profound weakness in the affected upper extremity. In rare cases, the main presenting symptom of this condition may be dyspnea on exertion secondary to an isolated unilateral brachial neuritis of the phrenic nerve.

Keywords: Parsonage-Turner syndrome; acute brachial neuritis/plexitis; isolated unilateral brachial neuritis; phrenic nerve; marathon runner; dyspnea on exertion

Parsonage-Turner syndrome, or acute brachial neuritis/plexitis, is a rare condition with an incidence of 2 cases per 100 000.¹ The cause of this disorder is unknown, but about a quarter of patients present with antecedent flulike symptoms.¹⁰ This syndrome typically affects the brachial plexus, with a characteristic pattern of severe, sharp neuropathic pain in the neck, shoulder, and arm, followed by profound weakness in the affected extremity. As weakness develops, the symptom of pain generally subsides. In a small percentage of cases, Parsonage-Turner syndrome involves isolated extrabrachial nerves, such as the phrenic nerve, which often results in dyspnea secondary to diaphragmatic paralysis or hemiparesis. A thorough review of the literature reveals only a small number of reported cases of bilateral diaphragmatic paralysis. As such, the present report is a rare case study of isolated unilateral brachial neuritis of the phrenic nerve in a marathon runner.

CASE REPORT

An otherwise healthy 49-year-old female competitive marathon runner presented to her primary care physician with worsening sore throat, dry cough, subjective fever, and left ear pain 8 days after running a marathon. She was diagnosed with bronchitis and treated with azithromycin. Fifteen days later, the patient returned to her primary care physician's office complaining of

dyspnea on exertion. She usually ran 5 to 15 miles per day but reported dyspnea after running less than 1 mile, with associated left-sided chest pressure with inspiration and exertion. The previous upper respiratory tract symptoms had resolved. The patient added that the dyspnea and chest pressure started when she woke up one morning with "terrible pain" in the left posterolateral neck area, radiating to her left shoulder. This pain symptom spontaneously resolved after a few weeks. The patient denied any weakness in the shoulder, arm, or hand.

During the second visit, the patient had normal vital signs as well as an oxygen saturation of 99% on room air. Physical examination findings were unremarkable. An electrocardiogram showed normal sinus rhythm with a lead II T wave inversion. Findings from the chest radiograph were interpreted as normal by the radiologist (Figure 1). Moreover, the complete blood count, thyroid-stimulating hormone, and D dimer levels were all normal. The patient's symptoms persisted despite treatment with an albuterol inhaler, fluticasone-salmeterol (inhaled), tiotropium (inhaled), and omeprazole. An exercise treadmill test yielded a negative result, and a flexible endoscopy did not demonstrate any vocal cord dysfunction. Finally, a high-resolution computed tomography scan was ordered secondary to a remote family history of interstitial lung disease and thus revealed an elevated left hemidiaphragm. A chest fluoroscopy, or sniff test (ie, a real-time radiograph that evaluates diaphragmatic movement), was

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Figure 1. The chest radiograph shows that the left hemidiaphragm is elevated compared to the right in full inspiration.

done and showed evidence of left hemidiaphragm paralysis (Figure 2). A subsequent nerve conduction study confirmed an absent left phrenic nerve response and a normal study on the right (Table 1).

Physical therapy was started to focus on inspiratory muscle strengthening and stretching, as well as joint mobilization for the ribs, cervical spine, and thoracic spine. In addition, focused and resisted breathing techniques were taught. The initial spirometer measurement was 1800 mL; after 4 months of consistent treatment, this increased to 2700 mL. The patient was able to gradually increase her running to 40 miles per week. Ten months after her initial presentation, the patient did achieve her goal of running a marathon without complications albeit slower than her typical times. Six months later, the patient completed another marathon with a personal best time.

Her spirometer readings were consistently in the range of 3100 to 3400 mL.

DISCUSSION

In 1897, Feinburg first illustrated a case report of unilateral brachial plexus neuritis associated with influenza.³ Fifty-one years later in the *Lancet*, Parsonage and Turner described 136 cases of this condition and coined the term *shoulder-girdle syndrome*, or neuralgic amyotrophy.⁹ This condition has a peak incidence between the third and fifth decades of life and is male predominant—with reports of 2:1 to 12:1.^{3,5,7,9,10} Moreover, this syndrome occurs bilaterally in about 33% of cases, but one side may be subclinical in presentation and detected only by evidence of axonal degeneration on electromyogram.^{2,7} To my knowledge, there has been only one other reported case of an isolated unilateral acute brachial neuritis of the phrenic nerve to account for dyspnea on exertion.⁶

Furthermore, electrodiagnostic testing is crucial to confirming the clinical diagnosis. In general, nerve conduction study and electromyogram findings are usually found 3 weeks after the onset of symptoms.⁴ In this case, the nerve conduction study demonstrated an absent left phrenic nerve response with a normal study on the right (table 1). However, a diaphragm electromyogram was not performed, because the neurologist thought that it would not add any further information and that the risks of the procedure outweighed the benefits. Therefore, there may have been subtle subclinical axonal degeneration on the right diaphragm that may not have been detected.

This case also illustrates the importance of obtaining a pulmonary function test when one suspects a pulmonary condition. A simple procedure, this diagnostic tool alone would likely have expedited this patient's treatment. Isolated brachial neuritis of the phrenic nerve can be suggested by pulmonary function tests that exhibit restrictive physiology and decreased inspiratory pressures with preserved functional residual capacity.⁵ Expiratory reserve volume and maximum expiratory pressure are generally preserved in isolated phrenic nerve palsy.⁵ Also note that findings from this patient's chest radiograph were originally interpreted as normal by the radiologist (Figure 1). In hindsight, upon review of the chest radiograph, the left hemidiaphragm is indeed elevated compared with the right in full inspiration.

The treatment of Parsonage-Turner syndrome is largely supportive, including analgesics, physical therapy, and patient education. Approximately 90% of patients attain a complete recovery in strength at 3 years after presentation.¹⁰ However, other studies have reported that almost half of patients have some residual deficit at long-term evaluation.⁸ During initial workup, routine laboratory tests should be done to rule out common causes of peripheral neuropathy, such as autoimmune diseases, diabetes, vasculitis, and infection.⁵ Tsairis et al found that the administration of corticosteroids reduces pain but does not appear to improve time to functional recovery.¹⁰ Moreover,

Figure 2. A chest fluoroscopy, or sniff test. Time passes in approximately 10-second intervals with each downward frame. The blue dotted line represents the right and left hemidiaphragms at rest in full expiration. The yellow dotted line represents each hemidiaphragm as it moves from full expiration to inspiration and back to expiration. A, the right hemidiaphragm demonstrates a full contraction as it moves caudally with deep inspiration (yellow arrow in the third frame); B, the left hemidiaphragm exhibits only some minimal flattening of the diaphragm (again, the yellow arrow in the third frame). Therefore, the sniff test was positive for left hemidiaphragmatic paralysis.

Table 1. Nerve conduction study: Supraclavicular fossa.^a

	Phrenic Nerve	
	Right	Left
Latency, ms	7.1	No response
Amplitude, mV	0.6	No response
Diaphragm		
Latency difference, ms	7.1	0.0
Distance, mm	260	0.0

^aNerve conduction study shows an absent left phrenic nerve response and a normal study on the right, thus confirming the cause of the left hemidiaphragmatic paralysis secondary to an isolated left phrenic nerve palsy.

anecdotal reports of success with plasma exchange and intravenous immunoglobulin have been recorded.⁵ Nearly 11 months after the onset of symptoms, our patient achieved her

goal of completing another marathon. As this case illustrates, obtaining a thorough clinical history about preceding pain, weakness, and other sensory symptoms helps lead one to an appropriate diagnosis and avoid unnecessary studies.

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