

Case Report Yonsei Med J 2021 Sep;62(9):868-871 https://doi.org/10.3349/ymj.2021.62.9.868



Parsonage-Turner Syndrome Following Typhoid Vaccination

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Parsonage-Turner syndrome is a rare neurological disease of varying etiology characterized by severe shoulder pain, muscle weakness, and atrophy. Mechanisms are unclear, but are thought to be genetic and immune-mediated reactions. Rarely, Parsonage-Turner syndrome occurs as a side effect of vaccination. A 20-year-old male who worked as a soldier visited the military hospital because of shoulder pain after vaccination against typhoid and was diagnosed with Parsonage-Turner syndrome based on electromyography and joint magnetic resonance imaging. Pain was controlled with a nerve block. Intravenous immunoglobulin was administered for improvement of neurologic symptoms. This case suggests that Parsonage-Turner syndrome should be considered as a side effect of vaccination. To the best of our knowledge, this is the first report of Parsonage-Turner syndrome following vaccination in Korea.

Key Words: Parsonage-Turner syndrome, brachial plexus neuritis, typhoid vaccine, vaccination, intravenous immunoglobulins

INTRODUCTION

Parsonage-Turner syndrome is a rare peripheral neurological disorder characterized by acute and severe neuralgia of the shoulder. After a sudden onset of pain, upper extremity muscle strength weakness and muscular atrophy occur. Pain usually improves in days to weeks, but muscle weakness can last for months to years. Causes of Parsonage-Turner syndrome include genetics, infection, vaccination, surgery, anesthesia, rheumatoid disease, trauma, radical exercise, and radiation therapy. The basic treatment is conservative, and the natural prognosis is good.

Received: August 12, 2020 **Revised:** June 15, 2021 **Accepted:** July 13, 2021

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• The authors have no potential conflicts of interest to disclose.

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This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (https://creativecommons.org/licenses/ by-nc/4.0) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited. Typhoid fever is a systemic disease accompanied by high fever and gastrointestinal symptoms caused by *Salmonella typhi*. In Korea, the incidence rate of typhoid fever has decreased, although it remains a concern worldwide. Typhoid vaccination is recommended before traveling to areas endemic for typhoid fever, after coming in contact with a typhoid patient, and when a pandemic occurs. Side effects of typhoid vaccines are the same as those of other common vaccines. Unusual, but severe, side effects include anaphylaxis and Guillain-Barré syndrome. However, it is rare for patients to develop Parsonage-Turner syndrome as a side effect of vaccination, although the exact incidence thereof is not known.

Here, we report a case of Parsonage-Turner syndrome in a patient after vaccination against typhoid fever along with a literature review.

CASE REPORT

A 20-year-old male visited our hospital because of pain in his left shoulder for 3 days. There was no relevant past or family history, including no history of side effects of vaccination, trauma, or viral illness. The patient was vaccinated against typhoid fever 6 days before in the left shoulder deltoid muscle, and there had been no other abnormal reactions. Vital signs were as follows: blood pressure, 127/82 mm Hg; pulse rate, 73 beats/ min; respiratory rate, 16 breaths/min; and body temperature, 36.9°C. The patient complained of pain around the left shoulder joint. The pain intensity was reported as a visual analog scale (VAS) score of 9.

During physical examination, no heat sensation, edema, or direct tenderness was observed at the injection site. Passive range of motion of the left shoulder joint was limited (Table 1). The upper extremity muscle tone and deep tendon reflexes were normal.

Peripheral blood tests showed no unusual findings other than mild increased liver function: aspartate aminotransferase, 44 IU/L; alanine transaminase, 112 IU/L. There were no abnormal findings in the left shoulder joint and surrounding bone structure on simple X-ray. A mild atrophic change of the infraspinatus muscle was identified on shoulder magnetic resonance imaging (MRI) performed at the time of the visit (Fig. 1).

To examine sudden joint pain, muscle weakness, and atro-

Table 1. Manual Muscle Test

Day	Shoulder abduction	Shoulder flexion	Elbow flexion
At visit	0	0	3
IVIG start D+1	0	0	3+
IVIG start D+3	1	1	4
IVIG start D+6	2	2	5
IVIG start D+9	2+	2	5
IVIG start D+12	2+	2+	5
IVIG start D+20	4	4	5

Grade 0, zero; 1, trace; 2, poor; 3, fair; 4, good; 5, complete. IVIG, intravenous immunoglobulin.



Fig. 1. Increased signal intensity with an ill-defined margin around the infraspinatus tendon and at the adjacent subchondral bone of the humeral head (A and B fat-suppressed T2-weighted saggital images, arrow, C and D fat-suppressed T2-weighted transverse images, arrowhead).

Table 2. EMG and Nerve Conduction Velocity

				Motor side	e to side c	omparison	table				
Nomo	Colored and	Recording -		LatOn (ms)		B-P Amp (mV)		Dist (mm)		CV (m/s)	
Nerve	Stimulus			L	R	L	R	L	R	L	R
Median	Wrist	APB		3.10	3.47	9.51	7.55			n/a	n/a
Elbow				6.80	7.43	7.15	6.43	210	260	56.8	65.5
Ulnar	Wrist	ADM		2.33	3.13	11.09	12.37			n/a	n/a
	B. Elbow			5.57	6.17	10.10	10.03	220	220	68.0	72.5
Radial	Forearm	EIP		2.13	2.20	4.75	5.88			n/a	n/a
	Elbow			6.13	6.63	3.09	1.80	250	260	62.5	58.6
Musculo-cutaneous	Erb's	Biceps		4.90	3.90	1.56	6.11			n/a	n/a
Axillary	Erb's	Delto	bid	6.67	3.43	0.23	13.12			n/a	n/a
Needle EMG											
Side	Muscle	Ins. Act.	PSW	Fibs.	CRD	Fascics.	MU Amp.	MU Dur.	Polyph	Recruitment	Cooperation
	Deltoid	Ν	2+	2+	Ν	Ν	Ν	Ν	Ν	No	Ν
	Biceps Brachi.	Ν	Ν	Ν	Ν	Ν	Ν	Ν	Inc	Discrete	Ν
	Supra-spinatus	Ν	3+	3+	Ν	Ν	Ν	Ν	Ν	Ν	Ν
	Infra-spinatus	Ν	3+	3+	Ν	Ν	Ν	Ν	Ν	Ν	Ν
Left	FCR						Ν				
	APB						Ν				
	ADM						Ν				
	Dors. Int. 1	Inc	Ν	Ν	Ν	Ν	Ν	Ν	Ν	Reduced	Ν
	Default						Ν				

EMG, electromyography; L, left; R, right; LatOn, onset latency; B-P Amp, baseline-peak amplitude; Dist, distance; CV, conduction velocity; B. Elbow, below elbow; APB, abductor pollicis brevis; ADM, abductor digiti minimi; EIP, extensor indicis proprius; Ins. Act., insertional activity; PSW, positive sharp wave; Fibs, fibrillation; CRD, complex repetitive discharges; Fascics., fasciculation; MU Amp., motor unit amplitude; MU Dur., motor unit duration; Polyph, polyphase; Inc, increased; FCR, flexor carpi radialis; Dors. Int. 1, first dorsal interossei; N, normal; n/a, not applicable.

phy, we conducted electromyography (EMG) and nerve conduction velocity for suspected Parsonage-Turner syndrome (Table 2). The results of the test revealed left upper trunk brachial plexopathy, which was diagnosed as Parsonage-Turner syndrome.

We initially administered a nerve block in the left C5 and upper brachial plexus using triamcinolone and lidocaine for pain control. Subsequently, pain decreased to 1 to 2 points on VAS. Initially, although steroids are usually available, we decided to administer intravenous immunoglobulin in consideration of autoimmune-related reactions. Intravenous immunoglobulin was administered for 5 days at a dose of 400 mg/kg/ day as a treatment for neuropathy. There were no side effects during the immunoglobulin administration. From the fourth day of immunoglobulin administration, the abduction and adduction ranges of shoulder joint motion increased, and muscle strength began to improve (Table 1).

The patient was discharged with complete improvement in muscle strength without pain complaints on hospital day 21. He was also followed-up at our outpatient department a month after he was discharged from the hospital. Recurrence has not been observed.

DISCUSSION

Parsonage-Turner syndrome was first reported in The Lancet in 1948 by Parsonage and Turner.¹ The prevalence of this syndrome is low. One study reported 1.64 cases per 100000 people.² There appears to be a male predominance, with a maleto-female ratio between 2:1 and 4:1.³

The causes of Parsonage-Turner syndrome vary. Parsonage-Turner syndrome occurring after vaccination was first reported by Rigal, et al.⁴ in 1956. Several cases of Parsonage-Turner syndrome caused by the polio vaccine, chickenpox vaccine, hepatitis B vaccine, influenza vaccine, and human papillomavirus vaccine have also been reported, but to date, no cases of Parsonage-Turner syndrome have been reported after typhoid vaccination.

The pathophysiological mechanism of Parsonage-Turner syndrome is unclear. The cause of the disease is thought to be autoimmune-associated neuropathy, and genetic factors also seem to affect the occurrence. Overall, Parsonage-Turner syndrome is thought to be of autoimmune origin, and a few studies have attempted to elucidate its pathophysiology.^{5,6} Vaccination-induced Parsonage-Turner syndrome may be associated with autoimmunity, but this is unclear.

Parsonage-Turner syndrome is a clinical diagnosis. EMG is a diagnostic test that can better identify and grade the severity of



denervation and re-innervation of the involved muscles. The MRI findings of Parsonage-Turner syndrome are thought to reflect denervation injury.⁷

There is no specific treatment for Parsonage-Turner syndrome. The basic management is conservative. Physical therapy may be helpful for preserving the range of motion but does not hasten recovery. During the onset of symptoms, the administration of oral prednisolone could shorten the duration of pain and accelerate recovery in some patients.8 In 2009, a Cochrane review identified only one open-label, retrospective series, the results of which suggested that administration of oral prednisolone during the first month of an attack could shorten the duration of painful symptoms and accelerate recovery in some patients.9 One study reported cases of immunoglobulin with methylprednisolone administration.¹⁰ However, some studies have reported that intravenous immunoglobulin (IVIG) use is effective for Parsonage-Turner syndrome in which treatment with steroids has failed, especially during acute periods. We used IVIG as a primary treatment, judging that, although it is an expensive drug, it is excellent in terms of effectiveness.^{11,12}

To the best of our knowledge, this is the first study describing Parsonage-Turner syndrome following vaccination in Korea. In rare cases, Parsonage-Turner syndrome should be identified when assessing side effects after vaccination, and immunoglobulin administration should be considered when it occurs.

AUTHOR CONTRIBUTIONS

Conceptualization: Jeong-Gil Kim and Dong Ho Jo. Data curation: Se Yong Kim. Investigation: Dong Ho Jo. Methodology: Dong Ho Jo. Project administration: Hong Sang Oh. Resources: Jeong-Gil Kim. Supervision: Hong Sang Oh. Validation: Dong Ho Jo. Visualization: Se Yong Kim. Writing—original draft: Jeong-Gil Kim. Writing—review & editing: Dong Ho Jo. Approval of final manuscript: all authors.

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