

Tetanus Presented as Hemiparesis and Facial Dystonia

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Abstract

We report a case of tetanus presenting as the facial dystonia and hemiparesis with complete recovery and video accompanied follow-up. A 78-year-old male, who sustained an abrasion to his right forearm, presented with dysarthria, dysphagia, prominent involuntary painful muscle spasms in the face, blepharospasm, risus sardonicus, trismus (lockjaw), neck muscle rigidity and mild weakness of the distal right upper extremity. The combined treatment of human antitetanus immunoglobulin, tetanus vaccine, penicillin, metronidazole and diazepam were given with no effect. He continued to worsen and due to the risk of the respiratory failure (finally not developed) he was sent to the Intensive Care Unit. The combined treatment was continued, and his symptoms started to improve within 2 weeks. As the muscle rigidity has been improving, the patient developed significant right hemiparesis. The imaging of the brain and cervical spine found no significant pathology. At the follow-up visits the patient presented only slightly impaired dexterity of the right hand that gradually resolved. There are many reports of unusual presentations of tetanus, however, hemiparesis is uncommon, and to our knowledge, only one case was reported in the literature. We highlight the variability of clinical presentation of tetanus, the need to consider tetanus in cases of rapidly evolving muscle spasms following peripheral injuries and provide the guidelines on tetanus prophylaxis.

Keywords: Tetanus; Lockjaw; Dystonia; Hemiparesis; Blepharospasm

Introduction

Tetanus is an infectious disease caused by *Clostridium tetani*, a gram positive, anaerobic bacillus, which produces spores that occur in the feces of many domestic animals, humans and in soil [1,2]. Once spores enter the body, they germinate and produce tetanus toxin which is internalized into nerves and spreads from the peripheral to the central nervous system by retrograde axonal transport, trans synaptic spread, and additionally by blood and the lymphatic vessels [1]. The toxin exerts its effect by preventing the release of gamma-aminobutyric acid (GABA) and glycine into the synapse and thus causes motor neurons to receive continuous excitatory discharge which result in the motor spasms [1].

Tetanus may occur as a result of a minor trauma and in up to 30% of cases no portal of entry is identified [1]. The disease is preventable and after the introduction of vaccinations its incidence decreased significantly, however, it still poses a significant health threat in the developing countries and still may occur in the developed countries, particularly among the elderly and other inadequately immunized people [1,3]. In 2013, 14 cases were reported in Poland, all of whom were over 60 years old, only 1 received a booster dose of tetanus, 5 received post-exposure prophylaxis, 7 died (50% mortality rate) [4]. The diagnosis of tetanus is clinical, and it is often delayed in areas, where the disease is uncommon [1,2]. Here we report a severe case of tetanus presenting as the facial dystonia and hemiparesis with complete recovery and video presentation at onset and follow-up.

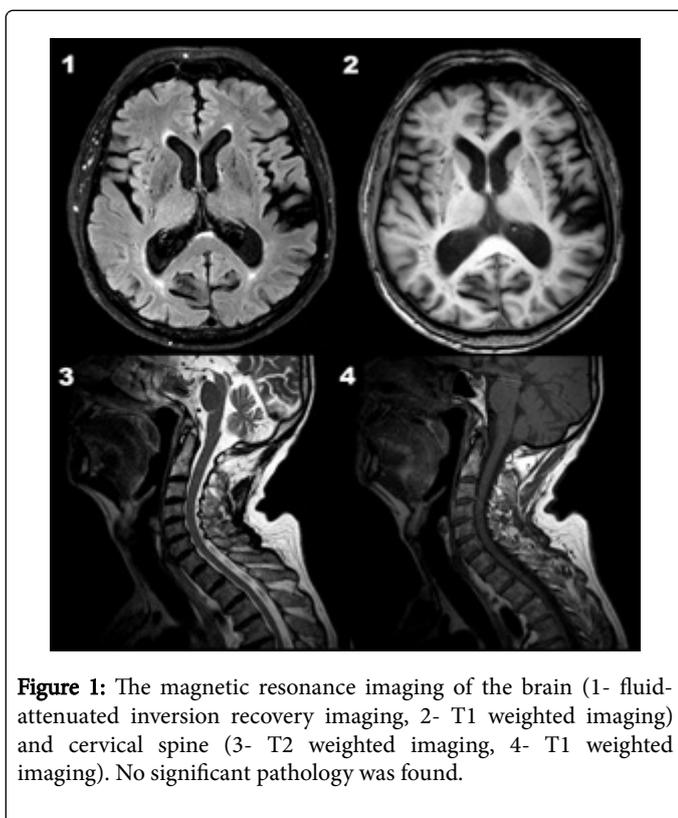
Case Report

A 78-year-old male, with a history of chronic renal disease, hypertension, hypothyroidism and paroxysmal atrial fibrillation, tripped and fell on the sidewalk, sustaining an abrasion to his right forearm. Five days after the accident he developed swallowing problems, followed by prominent involuntary painful muscle spasms in the face, articulation disturbances and weakness of the right upper extremity two days later. He was consulted by his family doctor, who referred him to the neurology outpatient department. The consulting neurologist suspected ulnar nerve injury and referred him to an orthopedic surgeon. Plain X-rays showed no abnormalities and he was then referred to neurology department. On the initial examination, 10 days after the accident, he presented with blepharospasm, risus sardonicus, trismus (lockjaw), dysarthria, dysphagia, neck muscle rigidity and mild weakness of the distal right upper extremity. The computed tomography of the brain was normal.

The clinical diagnosis of tetanus was made, and the patient was sent to the local Hospital for Infectious Diseases. On admission to the hospital the patient was given 6000 IU of human antitetanus immunoglobulin (Igantet) and tetanus vaccine (Tetana). The combined treatment of penicillin, metronidazole and diazepam was given as well. Due to the inflammatory changes at the site of the injury, aerobic and anaerobic cultures were taken. They were positive for methicillin-resistant *Staphylococcus aureus*, *Enterobacter kobei* and *Klebsiella oxytoca*, therefore ciprofloxacin was added. The patient continued to worsen and due to the risk of the respiratory failure (finally not developed) he was sent to the Intensive Care Unit at day 18th after the accident. The combined treatment was continued, and his symptoms started to improve within 2 weeks. As the muscle rigidity has been improving, the patient developed significant weakness of the right

upper extremity and mild weakness of the right lower extremity and was sent to the rehabilitation hospital. On admission he was wheelchair dependent and his Barthel Index of Activities of Daily Living (BI-ADL) was 8 points. After two months he was discharged home walking unassisted, with residual weakness of the right upper extremity and BI-ADL score of 20 points.

At the successive follow-up visits (10-28 months after the accident) the patient presented only slightly impaired dexterity of the right hand that gradually resolved. Electroneurography demonstrated axonal sensory right ulnar nerve neuropathy, while electromyography and repetitive nerve stimulation were normal. A 3-tesla magnetic resonance imaging of the brain and cervical spine showed no significant pathology (Figure 1).



Discussion

Tetanus may present as generalized (including neonatal) and localized (including cephalic) forms. The spectrum of symptoms is wide and includes rigidity, muscle spasms and dysautonomia that may lead to trismus (lockjaw), risus sardonius, opisthotonic posturing, dysphagia and respiratory difficulty [1,2,5]. Interestingly, there are many reports of unusual presentations of tetanus including blepharospasm, ptosis, ocular palsies, supranuclear opthalmoplegia, nystagmus, hemifacial spasm, cranial palsies, camptocormia and spastic paraplegia [6-11]. The pathophysiology underlying muscle paralysis in the course of tetanus is incompletely understood. Clinical and electrophysiological studies confirmed axonal polyneuropathy of “dying back” mode caused by tetanus toxin and was suggested in a series of 34 and 40 patients [12,13]. In a series of 25 patients reversible myopathic changes were found in the acute stage of tetanus [14]. However, hemiparesis is uncommon, and to our knowledge, only one

case was reported in the literature. In one of the reports the hemiparesis was attributed to lesions in the cortical and subcortical lesions in the frontal and parietal lobes followed by marked brain atrophy [15]. In our case imaging of the brain and cervical spine detected no lesions that could be responsible for the hemiparesis.

The diagnosis of tetanus is exclusively clinical. The spatula test (positive if severe spasms of the masseter muscles are elicited by touching the posterior pharyngeal wall) was demonstrated to be highly specific (100%) and sensitive (94%) for diagnosis of tetanus. Laboratory tests can demonstrate *Clostridium tetani* in 30% of patients, however, positive cultures were also found in immune individuals without the disease. Differential diagnosis of tetanus includes strychnine poisoning, orofacial infections, drug-induced acute dystonic reactions, mandibular fractures, progressive encephalomyelitis with rigidity and myoclonus (PERM), hypocalcemic tetany, rabies, Bell's palsy, stroke, serotonin and neuroleptic malignant syndrome, meningitis, epileptic seizures, psychogenic disorders. Most of the mimics can be discerned by a detailed history (recent injury, even if the presenting symptoms are remote) and physical examination, however, in some cases further investigations are required (e.g. toxicologic testing) [2,16-19].

According to World Health Organization guidelines, in the case of an injury, a dose of tetanus toxoid should be administered if the last dose was given more than 10 years ago or 5 years in the case of severe injuries [3]. Additionally, tetanus antitoxin may be used for prophylaxis in some patients e.g. insufficiently immunized [3].

Conclusion

In summary, tetanus diagnosis has to be considered in cases of rapidly evolving muscle spasms following peripheral injuries. Tetanus prophylaxis guidelines should be widely implemented as this rare complication may be life-threatening.

Ethical Publication Statement

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

Author Contributions

JD, MS, MD, JS worked on the manuscript.

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