

# Case Report

# Cephalic tetanus manifesting as isolated facial nerve palsy- a case report from rural Armenia

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#### Abstract

Cephalic tetanus is a rare clinical form of tetanus, clinically characterized by trismus and cranial nerve palsy involving one or more cranial nerves, facial nerve being the most common. We report a case of cephalic tetanus with left-sided lower motor facial nerve palsy in a 66-year-old non-immunized patient after an untreated laceration injury. The patient had dysphagia, spasm of the muscles of mastication, asymmetry of the left side of the face, cough, shortness of breath, and stiffness of neck muscles. The presentation was unique given that the facial nerve palsy appeared prior to the occurrence of trismus, which misled the initial diagnosis towards Bell's palsy. He was successfully treated with tetanus antitoxin without any adverse events. Although widespread use of tetanus vaccine has led to a dramatic decline in this fatal disease, sporadic disease occurrence is still possible, particularly in individuals without up-to-date vaccinations. In this case report we illustrate the importance of early recognition of cephalic tetanus prior to the development of the full clinical picture. The early initiation of therapy is the key to recovery from this deadly disease. Physicians are encouraged to include cephalic tetanus as a cause of facial nerve palsy in their differential. In particular, paying attention to cases manifesting early after head or neck injury.

Key words: Tetanus; facial nerve palsy; cephalic tetanus; cerebral tetanus.

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### Introduction

Tetanus is a non-communicable infectious disease caused by Clostridium tetani spores introduced through contaminated nonintact skin (cuts, wounds, abrasions, and lacerations) [1]. It is vaccine-preventable and if not timely treated, can be potentially fatal in those who are not immunized against the disease. About a million tetanus cases occur worldwide annually, and there are more than 200,000 deaths reported due to the disease [2]. The widespread use of the tetanus vaccine has led to a dramatic decline in this fatal disease [3]. For instance, tetanus caused 56,743 deaths worldwide in 2015, showing a decrease of 83% from 1990 during which there were 337,022 deaths [7]. However, the sporadic occurrence of the disease is still possible. This is particularly seen in individuals from developing countries where there are no up-to-date vaccinations, lack of proper adherence to the vaccination guidelines and documentation of immunization history [3]. Presently, about 90% of all tetanus-related deaths occur in developing countries like South Asia, Sub-Saharan Africa, or South-east Asia [7]. Other at-risk populations include older adults with waning immunity, diabetics, and intravenous drug abusers [8]. Four clinical forms of tetanus are recognized: generalized tetanus, neonatal tetanus, localized tetanus, and cephalic or cerebral tetanus [3]. Cephalic tetanus is one of the rarest clinical forms of tetanus, contributing to 0.9-3% of total cases and involves the neuromuscular system of the head and neck [4]. Cephalic tetanus commonly follows head injury, particularly at the face. Frequently, the route of the infection is the eye, however even the ear and tonsils can be the route of entry [5]. The characteristic feature of the Cephalic tetanus is the trismus in combination with the paralysis of one or more cranial nerves and/or ophthalmoplegia. Trismus usually precedes cranial

nerve palsies, facial nerve being the most common [5]. Depending on the cranial nerve involved, other clinical characteristics include neck stiffness, gaze deviation, risus sardonicus, and dysphagia. Complications include aspiration, respiratory paralysis, respiratory failure, and progression to generalized tetanus in approximately 66% of cases, with mortality of 15-30%[4]. The diagnosis of Cephalic tetanus is exclusively clinical and based on the patient's presentation. Trismus, resulting from the masseter muscle spasm, is frequently the first sign (in 50-75% of cases). This is usually followed by dysphagia, neck stiffness, and the signs and symptoms of cranial nerve palsy [10,11]. Cephalic tetanus with facial nerve palsy was first described by Rose in 1869 [6], but there are not many cases reported in the literature, particularly with facial nerve palsy preceding trismus. According to the WHO vaccine preventable diseases monitoring system- 2020 global summary, there have been no cases of neonatal or total tetanus cases in the last decade. There have also been no documented cases of cephalic tetanus from Armenia. But it is important to note that there has also not been a proper documentation of the current epidemiology of tetanus and patterns of mortality [7] Herein we report a case of cephalic tetanus from rural Armenia where trismus occurred after the facial nerve palsy and discuss with relevant literature review.

## **Case Report**

A 66-year-old Caucasian man presented to the emergency department at our center with complaints of inability to close the left eyelid and deviation of mouth to the right. The clinical history revealed a laceration injury to the mid-forehead due to an accidental fall upon a flower vase 17 days ago. He denied loss of consciousness, weakness of limbs, and fever following the incident and managed the wound at home without seeking medical care. Ten days after the injury, the patient experienced an inability to close his left eyelid and drooping of the left side of his mouth associated with difficulty chewing and drooling of saliva. With a provisional diagnosis of Bell's palsy, he was treated at a local health center by a neurologist with dexamethasone and valacyclovir for seven days, which did not improve his symptoms. It was then noted that the patient went on to develop a complete inability to open his jaw. Following this he was referred to our hospital for further evaluation and treatment.

At the time of admission, the patient had further developed dysphagia, spasm of the jaw muscles, asymmetry of the left side of the face, cough, shortness of breath, and stiffness of neck muscles. There was no fever, headache, vomiting, altered sensorium, or loss of consciousness. The wound examination revealed a partially healed 3×1 cm vertical laceration wound in the middle of the forehead. The rest of the neurological examination findings were normal. After the detailed clinical history, physical examination, unremarkable blood workup results, and normal CT scan, a clinical diagnosis of cephalic tetanus with left-sided lower motor facial nerve palsy was made. The history revealed that the patient was immunized against tetanus during his adolescence. The patient was hospitalized in the intensive care unit and treated with intramuscular administration of single-dose antitetanic serum 500 IU, intravenous metronidazole 500mg twice a day for ten days, supportive diazepam and eye care. The treatment course was uncomplicated. The patient was discharged on the 18th day of hospitalization with partially recovered facial nerve palsy, reduced frequency and severity of spasms and trismus, and recovered swallowing function. We counseled him about the disease and vaccination indications during the discharge. Follow-up at two months revealed complete resolution of trismus and some residual mouth deviation to the right side.

## Discussion

The Ministry of Health in Armenia recommends four doses of diphtheria-tetanus-pertussis. It is noteworthy that there have been no registered Tetanus cases by the United Nations Children's Fund (UNICEF)- Armenia, in the last decade. [9]. In the context of incidence of tetanus, we cannot exclude the possibility of undiagnosed and unreported cases, especially in the rural settings where there is no compliance with immunization guidelines, a lack of documentation of vaccination and lack of awareness in public about the disease. There has also not been a proper documentation of the current epidemiology of tetanus and patterns of mortality [7]. In our case, the patient was an older adult with possibly low immunity, who got infected with the pathogen via a lacerated wound [8]. Although about 66% of the patients with tetanus progress to generalized form with 15-30% mortality [4], we were able to successfully treat our patient probably because of early presentation and timely intervention. Given that Trismus is a key presenting sign in Tetanus our patient was diagnosed late despite that. The reason being that he first presented with facial palsy, before the trismus. It is uncommon for facial nerve palsy to precede trismus; for instance, there were only 5 cases found where trismus occurred after facial nerve palsy. One additional case was reported in

South Korea, in 2012 [12]. Meanwhile, the exact mechanism and the sequence of occurrence of cranial nerve palsies in tetanus are still to be determined [13]. It can also be a diagnostic dilemma when facial nerve palsy occurs before trismus, leading to consideration of differential diagnoses causing primary cranial nerve palsies such as Guillain-Barre syndrome, Miller Fischer syndrome, and myasthenia gravis. As the patient had a history of a recent open wound with no vaccination taken in the last ten years, had already developed trismus by the time he presented to us, and lacked weakness in any other body part and complained of fatigue, we could taper our diagnosis to tetanus. In this case report, we also illustrate the importance of early recognition of cephalic tetanus, one of the rarest forms, before the full clinical picture develops. The early initiation of therapy is the key to recovery from this deadly disease. As is in our case, a history of head or neck trauma is usually the mode of inoculating the responsible bacteremia into the body. This leads to a series of tetanus signs and symptoms. It is important to note that unlike other forms of tetanus that present with spasticity, the cephalic form presents with flaccid cranial nerve palsy [14], as is the case in our patient. Neurologists should be aware of cephalic tetanus as a cause of facial nerve palsy and should pay attention to cases manifesting early after head or neck injury. Physical examination findings can vary significantly from one patient to another [3]. Conditions such as strychnine poisoning, dystonia, seizure, dental infection, and hypocalcemic tetany should also be considered while suspecting tetanus in patients presenting with muscle spasms and trismus [3]. Nonspecific presentation of the disease may sometimes pose a diagnostic dilemma, where laboratory diagnostics and imaging are more useful for excluding other conditions rather than confirming tetanus. So, a high degree of vigilance and clinical suspicion, along with proper tracing of vaccination history in the settings of recent open wound injuries is imperative for the diagnosis of tetanus, which is almost always clinical [8,16]. Early diagnosis can guide treatment, including immediate hospitalization, administration of human tetanus immunoglobulin(intravenous immunoglobulin if unavailable), antispasmodics, antibiotics, wound care, tetanus toxoid booster, and sometimes, respiratory support [14].

## Conclusions

Tetanus cases are decreasing in the world but still common in developing countries with inadequate vaccination. Cephalic form, which is a rare form of tetanus, can be diagnostically challenging. Adherence to immunization guidelines and record-keeping can lower the incidence of such cases in the future, while strong clinical suspicion, meticulous history taking, and thorough exclusion of the possible alternative diagnosis can timely diagnose the disease. This can reduce disease morbidity, mortality, and health care burden.

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