Trismus as a Clinical Manifestation of Tetanus: A Case Report

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Although the incidence of tetanus disease has radically declined in developed countries, both dental practitioners and oral and maxillofacial surgeons should be knowledgeable about its diagnosis since initial manifestations of the disease, such as trismus and dysphagia, are observed in the orofacial region. This study reports on a case of generalized tetanus diagnosed in a middle-aged man. Before the tetanus diagnosis, the patient had sought medical advice from seven different health care professionals, including a dentist and an oral and maxillofacial surgeon. The patient reported trismus and dysphagia as his main complaints. The suspicion of tetanus emerged from the patient's manifestations in conjunction with his history of trauma and his agricultural occupation. The patient underwent successful treatment including administration of muscle relaxants, antibiotics, and booster vaccination doses of tetanus toxoid as well as a tracheostomy and aided mechanical ventilation. This case report highlights the significance of taking a meticulous medical history, thoroughly performing a physical examination, and systematically assessing orofacial signs and symptoms. J Oral Facial Pain Headache 2016;30:355–362. doi: 10.11607/ofph.1703

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etanus was first described as a clinical entity in ancient Greece by Hippocrates.¹ The word tetanus arises from the Greek verb "τείνειν" ("to stretch"), which describes muscle activity in the form of rigidity and spasms.¹ Nowadays, the incidence of tetanus has radically declined and the uncommon cases reported in the worldwide medical literature concern specific populations such as residents of developing countries and neonates.²⁻⁴ Epidemiologic studies in the United States, New Zealand, Australia, England, Wales, and Italy have shown that the majority of cases of tetanus in the developed world occur in aged individuals.⁵⁻⁸ In Greece, epidemiologic data from the Hellenic Center for Disease Control and Prevention (HCDCP) revealed that only 94 cases of tetanus were registered between 2000 and 2014.9 From 2001 to 2008, the average annual incidence of tetanus in the United States was 0.10 per 1 million in the overall population and 0.23 per 1 million among individuals aged 65 years or older; the fatality rate during that period was estimated to be 13%.10

Lack of immunization is considered to be the principal risk factor for contracting tetanus. In the United States, 75% of reported cases had not completed a primary immunization series.² The higher incidence of tetanus in the elderly is associated with low levels of the tetanus antibody in older age groups.^{11–17} In Greece, the guidelines of preventive vaccination against tetanus conform to those of the Advisory Committee on Immunization Practices (ACIP) and take place during early childhood and during military service of males.^{18,19}

The diagnosis of tetanus is based on clinical symptoms, with dysphagia, trismus, and neck stiffness being the earliest symptoms since the muscles of the jaws, face, and head and neck are first influenced as the tetanus toxin traverses along the shorter axonal pathways in this region in order to reach and affect the motor neurons located in the brainstem

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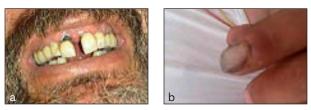


Fig 1 (a) The patient's active interincisal mouth opening of 18 mm. (b) The puncture wound in the patient's left hand.

or upper cervical spinal cord.²⁰ Painful spasms in the masseter and temporalis muscles cause trismus, or "lockjaw," while spasms of the pharyngeal muscles are responsible for dysphagia.²¹ Dental practitioners and oral and maxillofacial surgeons may be the first point of call for patients seeking medical advice because of restriction of mouth opening. Therefore, it is imperative for oral health care providers to be familiar with those pathologic entities that share trismus as a common sign. The etiology of trismus has been classified into two main categories, depending on an intraor extra-articular origin.^{22–52} It should be noted that the normal mouth opening range is between 40 mm and 60 mm (equivalent with two or three breadths of the index finger nail bed).⁵³

The primary aim of this report was to present a case of generalized tetanus diagnosed in the Department of Oral and Maxillofacial Surgery of Evaggelismos Hospital, Athens, Greece, and successfully managed in cooperation with other hospital specialists. A secondary aim was to focus on trismus not only as an early sign of tetanus, but also in terms of its differential diagnosis.

Case Report

A 53-year-old male sought medical advice at the Department of Oral and Maxillofacial Surgery, Evaggelismos General Hospital, Athens, Greece. The patient was mainly complaining of limitation in mouth opening, mild difficulty in swallowing food (dysphagia), a sore throat, slight photophobia, sound sensitivity, diffuse intraoral discomfort, and gradually exacerbated pain in the lumbar region. The patient reported that the symptoms had initiated 2 to 3 days before and progressively deteriorated. The patient was a farmer by occupation and lived in a rural town a few hundred kilometers outside of Athens. Between the onset of symptoms and his presentation to the Department, he had visited not only the local health center but also various health care specialists in Athens without receiving effective treatment. Specifically, the patient had previously requested

medical advice from a general practitioner, an internal pathologist, a dentist, an oral and maxillofacial surgeon, a neurologist, a general surgeon, and an otorhinolaryngologist. His medical history revealed diabetes mellitus (type 2) and long-term smoking (1 pack per day for 30 years). The most recent glycated hemoglobin (HbA1c) value was 7.2% (unregulated glycemic control). The information gathered in the dental history did not disclose either localized tooth pain or chronic or past disorders associated with the jaws, temporomandibular joint (TMJ), and masticatory muscles. In particular, before initiation of the symptoms, no report of episodic wide mouth opening and history of clicking was registered. No history of injury in the head and neck region was mentioned. The patient was hemodynamically stable with a normal level of consciousness, no fever, and normal resting breathing rate. The clinical examination revealed trismus with maximal interincisal opening (including a vertical overlap) of 18 mm (Fig 1a), painful spasms in bilateral masseter and temporalis muscles, mild neck stiffness, and lumbar rigidity. Clinical findings such as inflammation in the oral cavity, mandibular shift to the contralateral side, and cervicofacial swelling were absent. A localized edema on his left index finger (Fig 1b) was observed too, and the patient mentioned that he had suffered a puncture wound from a rusty nail during agricultural activities 9 days ago; the wound was left to heal without medical intervention. Radiologic examination included a chest x-ray, panoramic radiograph, and computed tomography (CT) scan of the head and neck. No pathologic finding was found, thereby reducing the possibility of trismus of intra-articular origin or originating from a facial fracture, or odontogenic infection.

Routine blood tests (including white blood cell count and C-reactive protein test) showed normal values, except for moderately elevated values of creatine kinase, creatine-kinase MB, and lactate dehydrogenase. Taking into account the inadequate immunization against Clostridium tetani reported in the patient's history, tetanus infection was suspected. Thus, the patient was admitted for observation and further investigation; he was made nil per os with intravenous administration of maintenance fluids. Lornoxicam 4 g together with orphenadrine citrate (muscle relaxant) 60 mg were intramuscularly delivered two times with a 5-hour interval in between. Despite the short-term relief for some of the symptoms, a few hours later, both trismus and lumbar rigidity were aggravated, and opisthotonos and extension of the lower extremities had developed. The spatula test was also performed and its outcome was positive.

A definitive diagnosis of tetanus was also clinically established and confirmed by an infectious disease specialist. The patient initially received 5,000 International Units of Human Tetanus Immunoglobulin (TETAGAM-P). Due to the impending laryngospasm and respiratory failure, this was immediately followed by tracheal intubation. The patient was transferred to the intensive care unit, where a tracheostomy was performed 5 days later. He remained in the unit for 32 days, 19 of which were spent in an isolated chamber to avoid any irritating visual and auditory stimuli. Antibiotic therapy for tetanus involved intravenous administration of metronidazole (500 mg \times 3) for 14 days. Propofol, midazolam, and muscle relaxants were given for the first 25 days and one booster injection of tetanus toxoid was administered on the 15th day. Approximately 4 weeks following the tracheostomy, assisted mechanical ventilation was replaced with controlled mechanical ventilation. Another booster injection was repeated on the 61st day of hospitalization. The patient was discharged in good clinical condition 63 days after his admission. During this long-lasting period, he developed several complications such as hospital-acquired pneumonia, thrombophlebitis of the right upper limb, malnutrition, decubitus ulcer in his lower back, urinary tract infection, and diskinesia. Several episodes of bradycardia also took place during the first 5 days of the patient's hospitalization. Eventually, all of these complications were appropriately managed.

Discussion

The causative factor of tetanus is *Clostridium tetani*, a Gram-positive, anaerobic, and spore-forming rod. The clinical features of a tetanus infection are attributed to the biologic effect of the neurotoxin tetanospasmin,⁵⁴ which enters the nervous system mainly at the myoneural junctions of the alpha motor neurons and follows an axonal retrograde transport to the central nervous system.²¹ Target cells of tetanospasmin include the glycinergic and the gamma-Aminobutyric acid (GABA-ergic) inhibitory neurons in the spinal cord and brainstem. The associated signs and symptoms occur as the inhibitory interneurons are affected and there is no inhibition of motor reflex responses to sensory stimulation. Failure of central inhibition results in a generalized tonic contraction of both agonist and antagonist muscles, which is the main characteristic of tetanic spasms.⁵⁵⁻⁵⁷ These muscular spasms are intensely painful and may be spontaneous or triggered by touch, visual, auditory, or emotional stimuli. In addition, if they are strong enough, bone fracture and/or tendon rupture may result.²¹

Since the diagnosis of tetanus is mainly clinically based, both history taking and clinical examination gain importance in the course of the disease. The prevalence of tetanus has largely been diminished in the Western world, and most practicing primary care physicians have never seen a single case in their career.⁵⁸ Therefore, it is possible that many oral health care providers may neglect or underestimate the early manifestations of tetanus infection. The patient in the present case sought medical care by seven different health care providers. None of them appropriately assessed the manifestations and all of them neglected the history of trauma in the patient's left hand. This tetanus case represents the second registered by oral and maxillofacial surgeons in Greece, with both trismus and dysphagia reported as the earliest manifestations of the disease.²⁵ Indeed, the majority of early complaints and presenting symptoms of tetanus are generally related to the head and neck region. The clinical presentation may vary and include one or more signs and symptoms such as trismus, dysphagia, alterations in facial movements, a scornful smile, speech disturbances, jaw pain, and dyspnea.⁵⁹ Trismus and dysphagia are provoked by the intensely painful muscle spasms of the masseter, temporalis, and pharyngeal muscles. Differential diagnosis should be made from a variety of commonly observed pathologic entities with similar clinical findings. Meticulous history taking in combination with proper physical, radiologic, and laboratory examinations are essential prerequisites to narrow the list of potential causes, and eventually lead to the correct diagnosis (Fig 2).

In the presented case, differential diagnosis between tetanus and a masticatory muscle disorder was reached after consideration of the (1) acute onset of signs and symptoms, (2) restriction in mouth opening which was becoming worse day by day, (3) severe restriction of maximum mouth opening, namely < 20 mm, (4) concurrent dysphagia, (5) bilaterally painful masseter muscle spasms, and (6) absence of a mandibular lateral shift. Based on the current taxonomic classification of masticatory muscle disorders, the clinical features of muscle pain (myalgia, tendonitis, myositis, and muscle spasm), muscle contracture, muscle neoplasms, and movement disorders can be associated with a restriction in mouth opening.^{60,61} If a patient presents with myofascial pain (ie, a subtype of myalgia), the limitation of movement may be slight, ranging from 35 to 45 mm, is unrelated to joint restriction, and is accompanied by unilateral and localized muscle tenderness that may be referred by trigger points (TrPs).³⁶ In cases of muscle spasm, according to the American Academy of Orofacial Pain, the diagnostic criteria encompass severe restriction in the range of motion and acute onset of pain at rest, which is aggravated by the function of the affected muscles. Electromyographic (EMG) activity is considerably higher at rest, while a sensation

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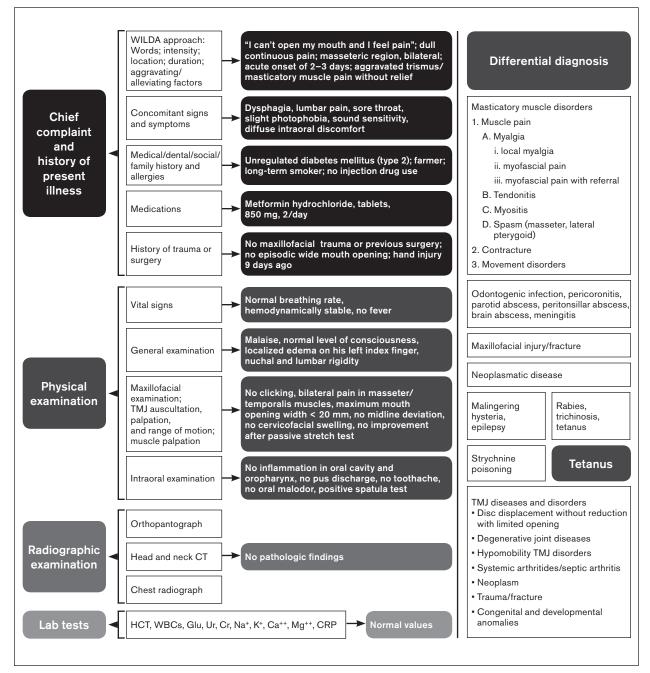


Fig 2 A diagram indicating the diagnostic progressive steps. CT = computed tomography; TMJ = temporomandibular joint; HCT = hematocrit; WBCs = white blood cells; Glu = glucose; Cr = creatinine; Ur = urea; CRP = C-reactive protein.

of muscle tightness or cramping may also be present simultaneously. Acute malocclusion or bite derangement may accompany the muscle spasm.⁶² When lateral pterygoid spasm exists, the mandible shifts to the contralateral side, leading to acute malocclusion, which is nevertheless reversible.⁶³

Myositis is unilateral muscle inflammation due to a spreading infection, external trauma, or excessive strain. Included in the signs and symptoms are constant and unilateral acute pain within the muscle, tenderness to palpation, edema, erythema, and/ or overlying increased temperature.³⁶ On the other hand, the muscle contracture may be a chronic condition, which would be characterized by continuous gross shortening of the muscle, little or no pain, a long-term history of trauma/infection/disuse, and a progressive decrease in range of motion.

TMJ disorders occurring with limited mandibular opening include disc dislocation without reduction and limited opening, hypomobility disorders

(ie, ankylosis), systemic arthritides (rheumatoid or psoriatic), degenerative joint diseases (ie, osteoarthritis), neoplasms, osteochondritis dissecans, synovial chondromatosis, condyle fractures, and congenital/developmental disorders (ie, coronoid hyperplasia).60,61 Joint locking due to disc displacement without reduction and limited opening manifests as mouth opening between 25 and 30 mm.⁴⁹ A previous history of clicking often coexists with an acute presentation of closed lock after an event such as biting into an apple. Mandibular shifts toward the affected joint during mouth opening may also occur.⁴⁹ Trismus may occur in septic arthritis and is accompanied by severe TMJ pain and acute malocclusion.⁵¹ Imaging studies contribute to a trismus diagnosis by demonstrating widening of the joint space and limitation of condylar movement. Magnetic resonance imaging of the TMJ is strongly recommended at the early stage of septic arthritis to allow for the detection of joint effusions.⁵¹ Computer tomography (CT) imaging is used in the chronic stage to image bony changes, articular space widening, cellulitis around the TMJ, and signs of any impending abnormalities such as infectious osteoarthritis, osteomyelitis, or ankylosis of the TMJ.⁵¹ TMJ hypomobility disorders, degenerative joint diseases, and congenital/developmental disorders can also be excluded by the radiologic and clinical examination. The passive stretch test is considered to be the principal diagnostic test for differentiating between a muscular cause of limited mandibular movement and intracapsular restriction. Among the techniques that have been described to perform the passive stretch test are spraying the masseter and temporalis muscles with a vapocoolant spray, and the maximum assisted opening of the jaw by applying force by the fingers between the maxillary and mandibular teeth.64

Trismus of odontogenic origin most commonly implies a pericoronal infection resulting from an erupting third molar. Both the patient's history and clinical examination may reveal acute onset of unilateral pain, inflammatory mucosa around the erupting tooth, unilateral facial swelling and/or redness, and fever.⁶⁵

A peritonsillar abscess is one of the nonodontogenic factors associated with trismus; it is typically characterized by fever, intense unilateral pain localized in the throat, dysphagia, redness and edema in the tonsillar area of the affected side, unilateral earache, and swelling of the jugulodigastric lymph nodes.²⁴ A parotid abscess usually exhibits swelling, erythema, and pain in the preauricular area unilaterally. Swelling is exacerbated during food intake while pus discharge can be noted clinically from the Stensen duct after palpating the swollen parotid gland. The patient's chief complaint may be oral malodor. CT imaging is indicated to detect a parotid abscess.²⁸ Meningitis is accompanied by a sudden and high fever, severe headache, neck stiffness, nausea or vomiting, confusion, difficulty in concentration, and seizures.²⁶ A brain abscess involves fever, headache, drowsiness, confusion, seizures, hemiparesis, and speech difficulties, and clinically progresses in a rapid pattern.²⁷

In cases of trismus as a result of facial skeletal trauma, the clinical findings can include pain, malocclusion, open bite, numbness, swelling, bruising, facial deformity, diplopia, or flattened malar eminence.^{30,31}

Trismus is rarely noted in cases of hydrophobia (rabies) and tetany.²⁹ The latter is characteristically differentiated by its carpopedal contractions. Trichinosis is a rare parasitic disease that can cause trismus in combination with high fever, periorbital edema, and eosinophilia.²⁹ This disease is acquired by eating raw or inadequately cooked infected pork (or other domestic and wild animals). In cases of strychnine poisoning, trismus develops after the establishment of generalized twitching and convulsions.²⁹ In epilepsy, the trismus lasts a short time, while hysterical patients exhibiting trismus may have convulsive seizures of various assorted forms.²⁹

No specific laboratory tests are currently available for routine use in the diagnosis of tetanus.⁶⁶ The spatula test is a clinical diagnostic means contributing to tetanus identification, and is made by touching the patient's posterior pharyngeal wall with a blunt instrument such as a wooden tongue depressor. Normally, a gag reflex is produced and the patient tends to expel the instrument (a negative test result). A positive test result implies involuntary spasm of the masseters and thereby the patient bites down on the "spatula." The test has shown high specificity (100%) and high sensitivity (94%) for diagnosing tetanus.⁶⁷

Since Clostridium tetani is an obligate anaerobic rod, tetanus-prone wounds are those with oxygen deficiency. As such, wounds being devitalized, crushed, and contaminated with dirt and rust as well as open fractures, punctures, and abscesses are characterized as tetanus-prone wounds.⁶⁸ The study by Bardenheier et al showed that 41% of the tetanus cases registered in the United States in 1987 and 1988 had been connected with gardening and farming activities.⁶⁹ The patient of the current report was a farmer and had a deep injury in his hand owing to a work accident. The greatest risk factor for contracting tetanus in the United States has been reported to be the lack of primary immunization.² Specific population groups including older adults, persons in poverty, men without military service, and patients with diabetes mellitus are susceptible to tetanus.^{70,71} The patient in the present case proved to have a history of diabetes with unregulated glycemic control

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before his hospitalization. There is evidence indicating that poor glycemic control is associated with an immunocompromised state.⁷² Hsu and Groleau have suggested both prophylactic use of Human Tetanus Immunoglobulin (HTIG) and frequent dosing of tetanus toxoid every 5 years in order to maintain sufficient "protective" antibody levels in immunocompromised patients.⁷⁰

Concerns should be also raised in immigration-heavy areas, including the United States and many European countries. The available scientific evidence has demonstrated that unauthorized immigrants and refugees lack adequate knowledge about the etiology, symptoms, transmission risk factors, prevention strategies, consequences, and treatment of infectious diseases such as Hepatitis B (HBV) and C (HCV) virus infections.73 Thus, it is possible that immigrants may not have received the essential tetanus immunization and/or ignore the appropriate preventive measures for wound management. The Tetanus Quick Stick (TQT) is a reliable, prompt, and cost-effective immunochromatographic test that detects anti-tetanus antibodies in human blood and can efficiently define immunity status of patients in an emergency department.74,75

Furthermore, a recent study by Petti et al investigated the awareness of dentists regarding preventive vaccination against infectious diseases including tetanus.⁷⁶ Of the 443 dentists participating in the survey, only 50 had received a tetanus toxoid booster dose during the past 10 years. This finding demonstrates that even health care providers misperceive or ignore the efficacy and benefit of tetanus vaccination. Additionally, this may reflect a possible inability of dentists to guide patients properly in terms of preventive vaccination for tetanus. Tetanus is a disease preventable by vaccine, so all health care providers should be able to evaluate their patients' vaccination status, especially if the patients are aged adults, injection drug users, diabetic, and/or suffer from chronic wounds.10

Conclusions

Tetanus is a rare but life-threatening infectious disease, and so its prompt diagnosis is considered mandatory. Provided that the most common initial manifestations of tetanus infection (ie, trismus, dysphagia, and neck stiffness) are localized in the head and neck region, both dental practitioners and oral and maxillofacial surgeons may be the first to encounter such cases. Thus, oral health care providers should effectively perform differential diagnosis from other commonly observed pathologic entities that share similar clinical findings, mainly trismus. This case report underlines the significance of meticulous history taking as well as the value of increased diagnostic awareness when evaluating signs and symptoms.

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Journal of Oral & Facial Pain and Headache 361

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