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Case report

# Unusual generalized tetanus evolving from odontogenic infection: A case report and review of recent literature



Muhammad Akbar<sup>a,\*</sup>, Muhammad Ruslin<sup>b</sup>, Andi Sitti Hajrah Yusuf<sup>b</sup>, Paolo Boffano<sup>c</sup>, Kei Tomihara<sup>d</sup>, Tymour Forouzanfar<sup>e</sup>

<sup>a</sup> Department of Neurology, Faculty of Medicine, Hasanuddin University, Makassar, Indonesia  
<sup>b</sup> Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Hasanuddin University, Makassar, Indonesia  
<sup>c</sup> Division of Dentistry, Vercelli Hospital, Vercelli, Italy  
<sup>d</sup> Division of Oral Maxillofacial Surgery, Niigata University Graduate School of Medical and Dental Sciences, Niigata City, Japan  
<sup>e</sup> Department of Oral and Maxillofacial Surgery, Oral Pathology and 3D Innovation Lab, Amsterdam University Medical Center, Vrije Universiteit Amsterdam, Amsterdam Movement Sciences, Amsterdam, the Netherlands

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## ABSTRACT

Tetanus is an acute and vaccine-preventable disease caused by anaerobic bacteria, *Clostridium tetani*. This bacterium can enter the human body via a deep wound, burn injury or medical procedure; however, certain cases also originate from odontogenic infection. In the present study, a tetanus infection associated with dental origin in a 44-year-old man is reported. The case was complicated by lockjaw and difficulty swallowing that worsened over a few days, followed by a generalized spasm. Furthermore, a literature review was performed, in which six reported cases of tetanus, presumed to be of dental or oral origin, were identified between 2011 and 2021. General practitioners, especially dentists, should be aware of tetanus associated with odontogenic origin even without a history of an external penetrating wound or other medical procedures.

## 1. Introduction

Tetanus is an acute and often fatal disease caused by anaerobic bacteria, *Clostridium tetani* [1, 2]. Although the incidence of tetanus has been declining over the past two decades due to immunization programs, it remains a major health problem in developing countries, with high mortality rates [1, 3]. In 2015, there were 56,743 deaths from tetanus worldwide [4]. In Indonesia, there were 3559 non-neonatal deaths, with a mortality rate of 1.38 per 100,000 persons [4]. The tetanus mortality rate is considered to be fairly high and therefore this infectious disease requires more attention [5].

One of the main diagnostic factors for tetanus is a history of recent penetrating wounds, internal wounds or surgical operations [1, 2]. In the absence of these factors, many physicians often do not consider tetanus as the differential diagnosis. To date, only a few studies have discussed the odontogenic origin of tetanus, with the presumed origin being a result of one of the following: tooth extraction, root canal therapy, injection of anesthetic, gross caries, periodontal abscess or intraoral soft tissue trauma [6, 7, 8, 9, 10, 11]. In the present case report, a case of generalized tetanus of presumed odontogenic origin is presented. Furthermore,

recent literature on tetanus caused by dental infection was reviewed. The present case report may be used to advise physicians and dentists and raise awareness concerning this often-fatal disease in the clinic.

## 2. Case report

A 44-year-old male visited the hospital due to his inability to open his mouth. He had initially been in good health; however, one week before being admitted, he experienced stiffness in his neck and jaw. He also complained of pain in the same region. These symptoms worsened over a few days and he had difficulty swallowing. The patient did not have a fever, ear infection, neck pain, neck trauma, or a history of an external perforated wound, and had not taken any medicine. He also denied a history of seizure or temporomandibular joint disorder. Related to the chief complaint, anamnesis revealed that the patient often poked his interdental gingiva with a toothpick. He had not been recently administered with a tetanus vaccinations and did not remember having any vaccinations when he was a child.

A general examination demonstrated that the patient's vital signs were normal. He had stiffness in his neck and trismus, with the maximum

<sup>2</sup>

\* Corresponding author.

E-mail address: [akbar@med.unhas.ac.id](mailto:akbar@med.unhas.ac.id) (M. Akbar).

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inter-incisal opening being 10 mm. His facial expression displayed risus sardonius. A routine blood test indicated elevated activity of hepatic enzyme aspartate aminotransferase (AST) to 107 U/L. The patient was consulted to the Oral and Maxillofacial Surgery Department and intraoral examination demonstrated that the patient had bad oral hygiene with some caries in his teeth. Dental caries were noted in the upper right quadrant of the mouth (teeth no. 11, 12, 15 and 16) and chronic gingivitis was also identified. The dental status of the patient was also evident via panoramic radiography. There was no visible swelling found in the neck and submandibular space.

The patient was diagnosed with tetanus and based on <sup>11</sup> Ablett classification [12] he was suffering from moderate disease. The patient was administered with a single intramuscular dose of 3000 IU human tetanus immunoglobulin (HTIG), 2 g intravenous ceftriaxone twice a day, 500 mg intravenous metronidazole three times a day, 5 mg midazolam/hour and 1 g paracetamol three times a day as needed. A nasogastric tube was placed to ensure fluid/feeding intake and prevent aspiration. Dental treatment was postponed until the trismus subsided.

While in the hospital, the patient felt stiff and rigidity in his abdomen and on the third day of admission he experienced a single episode of generalized muscle spasm. His blood pressure also fluctuated, ranging from 140/90 to 110/70 mmHg and a heart rate of 70 and 90. Within the following six days, the trismus gradually attenuated and the spasm did not occur. The patient opened his mouth to eat a week after treatment. After two weeks of hospitalization the patient was discharged.

The report was conducted according to established ethical guidelines and approved by the Research Ethics Committee of the Faculty of Dentistry Hasanuddin University with protocol number 0054/PL.09/KEPK FKG-RSGM UNHAS/2022. The patient's written informed consent was obtained for publication of this case report.

### 3. Discussion

<sup>24</sup> Tetanus is caused by the spore-forming bacterium *C. tetani* that is found in soil contaminated with animal and human excreta under anaerobic conditions [1, 2, 3]. *C. tetani* secreted two toxins: tetanospasmin and tetanolysin [13]. Tetanolysin can locally damage viable tissue surrounding the infection and optimize the conditions for bacterial multiplication [13]. Tetanospasmin is a potent neurotoxin that causes skeletal muscle contraction or autonomic dysfunction [3, 5, 13]. It is generally known that trismus is the main characteristic and initial sign of tetanus before progressing to generalized spasm [2, 11]. Aside from trismus or lockjaw, <sup>7</sup> common early indication of tetanus in older children and adults is abdominal rigidity and generalized spasms that are frequently induced via sensory stimuli [14, 15, 16]. In the present report, it was demonstrated that the patient's initial sign and symptom of suspected tetanus was trismus, followed by neck stiffness and risus sardonius, as well as a stiff and rigid abdomen. The patient's condition progressed and he experienced a generalized spasm, but fortunately his symptoms improved. The patient also exhibited mildly elevated AST activity, which was possibly due to muscle fiber damage from prolonged muscle contraction.

In the present report <sup>27</sup> tetanus diagnosis was determined using clinical results alongside the standard case definitions available from the World Health Organization (WHO) official website [17]. The WHO states that non-neonatal tetanus is defined by either a prolonged facial spasm that makes the individual appear to be grinning or painful muscle spasms [17]. In line with this, in 2009, the Council of State and Territorial Epidemiologists published a case definition of tetanus as an acute illness with muscle spasms or hypertonia in the absence of a more likely diagnosis [18]. According to the US Centers for Disease Control and Prevention, tetanus is a clinical condition with <sup>16</sup> conclusive laboratory tests [19]. The clinical definition of tetanus is characterized by painful muscular contractions, particularly of the masseter and neck muscles, and secondarily of the trunk muscles [19]. Supporting these aforementioned definitions, previously published studies have underlined that

tetanus can be defined as clinical evidence of illness without other apparent medical cause, with or without isolation of *C. tetani* and with or without history of injury [11, 14, 15, 20, 21].

In the case of the patient in the present report, the result of the bacterial culture for *C. tetani* was not used as a basis to <sup>28</sup> diagnose tetanus, particularly since the patient presented with indicative clinical signs and symptoms of <sup>25</sup> tetanus with no history of immunization. It has previously been agreed that the <sup>3</sup> diagnosis of tetanus should be based on clinical presentation and that treatment must not be delayed once isolation, culturing of the organism [5, 11, 22, 23]. Wound swabs for microscopy, culture and sensitivity tests may be performed, but the result may not strongly indicate the <sup>3</sup> presence or absence of the organism [11, 18, 20]. Although *C. tetani* is among the easiest bacteria to identify once isolated, culturing them from surgical material or wounds often presents certain difficulties due to the paucity of organisms usually present in wounds [6, 14, 16]. Culturing of *C. tetani* is frequently unsuccessful, with cultures only being successful 30% of the time, and negative results <sup>11</sup> do not eliminate the diagnosis [8, 11, 14, 18]. Moreover, a positive culture can be found in both people who have and do not have tetanus and this result does not indicate whether the organism contains the toxin-producing plasmid [8, 11, 18].

Tetanus of odontogenic origin is rare but has been reported in a few previous studies. To the best of our knowledge, only six cases of tetanus associated with odontogenic infection have been reported in the last decade. In Table 1, several cases of the odontogenic origin of tetanus caused by dental procedures, such as tooth extraction, root canal therapy, or injection of an anesthetic <sup>10</sup> are presented [7, 8, 9]. Moreover, other cases have also reported gross caries, periodontal abscess, or intraoral soft tissue trauma as presumed origin [6]. In one single-center retrospective study of 17 tetanus cases in China, Fan et al reported that the maxillofacial region was the point of entry of *C. tetani* in 5.9% of all cases [24]. In the present study, the entry point of *C. tetani* was suspected to be via dental <sup>4</sup> infection. This was suggested as no other lesions were apparent, there was no history of any cut, puncture wound, splinter or other such portal of entry, and the physical examination also did not reveal any such evidence. This suspicion was strengthened by the patient's history, whereby the patient often used a toothpick to poke his interdental gingiva, which might have predisposed him to the oral wound. A previously published study reported that when the muscles of the jaw and neck are involved, tetanus should be at the top of the differential diagnosis [23]. The results of the present study also support a previous report, whereby it was established that based on a patient's history, incomplete immunization status, <sup>5</sup> and in the setting of an odontogenic infection with a triad of rigidity, a clinical diagnosis of tetanus can be made [8, 11].

In the present report, the patient was treated with a single intramuscular dose of 3000 IU HTIG as the neutralization of unbound toxin is performed by the administration of HTIG or equine tetanus <sup>12</sup> immunoglobulin [5]. Furthermore, the patient was also treated with 500 mg intravenous metronidazole three times a day. Metronidazole is the main antimicrobial therapeutic for anaerobic bacteria, such as *C. tetani*. Its use is supported by an open-label study of 173 patients in Indonesia whereby 500 mg metronidazole was reported to the lower mortality rate from 24% to 7% when administered orally every 6 h [13]. In the present study, broad-spectrum antibiotics, including ceftriaxone, were also used to treat the patient as infections in the oral region are often polymicrobial.

There are certain principles in the management of tetanus, including wound care at the point of entry, antimicrobial treatment for *C. tetani*, neutralization of unbound toxin and symptomatic treatment, that address muscular spasm and autonomic dysfunction [1, 15]. For an external wound, care should be taken to debride and clean the wound to ensure aerobic condition; however, concerning odontogenic infection, treatment of dental infection can be challenging due to trismus. In the present study, owing to the maximum inter-incisal opening of 10 mm, the dentist decided to postpone dental treatments until the patient's condition improved.

**Table 1.** Review of literature published case reports of tetanus presumed dental or oral origin.

References	Country	Patient's Age	Suspected point of entry	Clinical manifestation	Bacterial culture	Diagnosis	Outcome
Baviskar et al. [6]	Mumbai	27 years old	Local anesthetic injection for carious left maxillary second premolar.	Trismus, dysphagia, neck and back rigidity, generalized spasm.	Negative culture for <i>C. tetani</i>	Tetanus	Death
Bernardes et al. [7]	USA	77 years old	Tongue ulcer	Trismus, generalized spasm, ptosis, ophthalmoparesis.	None	Cephalic Tetanus	Death
Darraj et al. [8]	Canada	42 years old	Multiple dental caries, recent tooth fracture of right upper third molar site.	Trismus, dysphagia, spasm of neck, shoulder, arms, back, and legs	Negative culture for <i>C. tetani</i>	Generalized tetanus	Discharged
Ulfa and Husna [9]	Indonesia	42 years old	Dental cavities	Trismus, dysphagia, rigidity.	None	Generalized tetanus	Discharged
Ajayi and Obimakind [10]	Nigeria	22 years old	Tooth extraction	intermittent spasm of the muscles of the face and neck.	None	Cephalic tetanus	Discharged
Bassey et al. [11]	Nigeria	70 years old	Tooth extraction under suspicious hygienic circumstances	visible muscular spasms	None	Cephalic tetanus	Died

The present report emphasized the need to recognize early clinical symptoms of tetanus and initiate effective treatment, and also highlighted the importance of immunization. Although tetanus infections from sources in the intraoral region are rare, practitioners must be aware of the possibility of their occurrence since this disease has potential dangers and is associated with a high mortality rate [2, 8, 11]. It is recommended that patients with a high risk are identified, the clinical manifestations of tetanus are recognized and cases are promptly referred to the appropriate department to ensure a better prognosis. Since the case in the present study is rare and there are numerous questions concerning the clinical diagnosis and laboratory testing of tetanus to be addressed, a systematic review on this subject will be important.

#### 4. Conclusion

In summary, oral mucous or teeth can be an entry point for *C. tetani*. Hence, general practitioners, especially dentists, should be aware of tetanus associated with odontogenic origin in the clinic. This infectious disease should always be considered in the differential diagnosis of a patient with trismus and muscle spasm in the neck, even without an external penetrating wound or medical procedure.

#### 15 Declarations

##### Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

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##### Data availability statement

Data will be made available on request.

##### Declaration of interest's statement

The authors declare no conflict of interest.

##### Additional information

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