

LETTER TO THE EDITOR

A CASE OF CEPHALIC TETANUS IN A DEVELOPED COUNTRY

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Tetanus is a potentially life-threatening infection that results from contamination of skin wound by *Clostridium Tetani* spores. Although, it remains an important health problem in developing countries without strict national immunization programs, this condition is rare in the developed world. The most frequent presenting symptoms are trismus and dysphagia, due to the spasmodic contraction of the masticatory muscles. Then the disease usually diffuses with a descending pattern inducing a generalized contraction of the agonist and antagonistic muscles, which characterize a tetanic spasm. Mortality usually results from respiratory failure, cardiovascular collapse, or autonomic dysfunctions. Treatment usually requires the prompt admission to the intensive care unit to avoid the development of potential life-threatening complications. We report the case of a 78-year-old farmer, who was referred to us with progressive onset of lock-jaw and muscular stiffness of the facial region, that had occurred after he had scratched himself with a rose. The recognition of the presenting signs of cephalic tetanus allowed the prompt management of the infection. However, because of the rarity of this condition, the clinicians may be unfamiliar with the clinical presentation, and be unsuspecting of the diagnosis.

Tetanus is potential life-threatening infection caused by the effects of the exotoxin from *Clostridium tetani* on the central nervous system.

Since the implementation of vaccination programs during the second half of the 20th century its incidence is globally declining. However, it continues to be a significant health problem and is associated with a high mortality rate, particularly in the developing countries with hot climates and densely populated areas rich in organic matter (1-3). The worldwide incidence of tetanus is estimated at 1 million cases annually (1). According to recent observations, the fatality rate of tetanus ranges from 11% to 70% depending on the availability of resources and intensive care unit to manage the disease (4, 5).

In developed countries the majority of cases

occur in the elderly (over 60 years of age) whose tetanus antibodies had declined with time, immunocompromised individuals and in intravenous users of contaminated drugs (6-10). In Italy, since the introduction in 1950 of mandatory neonatal vaccination schedules, a decrease of about 86% has been observed of the incidence of tetanus. In 2005, there were just 49 documented cases, and the last neonatal infection was reported in 1982 (11). Systemic tetanus is the most common manifestation of the disease, and in this condition, intensive care management has demonstrated to be effective in reducing the mortality rate by less than 30% (6). The most frequent presenting symptoms are trismus and dysphagia. However, because of the rarity of this condition, the clinician may be unfamiliar with the clinical presentation, and be unsuspecting of the

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diagnosis (12).

This study describes a case of timely diagnosed cephalic tetanus, which was diagnosed at an early stage of disease.

Case report

A 78-year-old man was referred to the Unit of Maxillofacial Surgery, University Hospital of Padova, Italy, in 2009 for the progressive onset of trismus and dysphagia associated with difficulty in speech. The previous day, he had gone to the Emergency Room of his city hospital, where he was discharged without diagnosis and exhorted to consult an oral and maxillofacial specialist.

The patient's medical history included only a symptomatic episode of gastric ulcer ten years previously and no other significant comorbid conditions. The patient reported that he had scratched his arm on a rose thorn approximately a fortnight previously, but he had not shown the wound to a doctor.

Initial evaluation showed a well-nourished man, who was alert and oriented to his surroundings. Oral examination showed a closed lock-jaw associated to bilateral masseter, and temporal and pterygoid muscle spasm. In detail, both intraoral and extraoral palpation revealed stiffness of masticatory muscles and advocated severe local pain. Forced mouth opening was identified with a metric ruler for linear measurement and it was approximately 1.5 cm; while spontaneous mouth opening was not attainable. Trismus and dysphagia had significantly developed and progressed in the previous 24 hours, and speech had also become difficult. Moreover, the patient presented risus sardonicus and photophobia, associated to initial spasm of the extensors of the neck and bilateral shoulders muscles stiffness.

Direct stimulation with a pallet of the oropharyngeal posterior wall (Spatula test) was restricted because of the limited mouth opening, but it resulted positive. Vital signs and serum biomarkers were recorded: blood pressure 160/80 mmHg, heart rate 80 beats/min, oxygen saturation on room air 98%, and body temperature less than 37°C. The white blood cell count was 5.150/ml, haemoglobin was 13.7 g/L and platelets were 234.000/mm³. All serum electrolytes were within normal range. The dosage of tetanus circulating antibodies was found

inferior to the protective value (0.03 IU/l).

The patient underwent panoramic radiography that showed compromised teeth and periodontal disease, but no signs of dental abscesses (Fig 1). A computed tomography was also performed that did not show either signs of evident odontogenic infections, or mandible fractures, or temporomandibular joint disorders (ankylosis, arthritis or disk pathology).

A diagnosis of tetanus was proposed and the patient was admitted for the prompt appropriate treatment. We administered the recommended intramuscular dose of tetanus immune globulin (500 IU). The patient was also given a 15-day cycle of intravenous antibiotics (ampicillin and sulbactam, 3 g tid; associated with metronidazole, 500 mg tid). The wound was also surgically debrided and treated daily. An anaesthesiological consultation excluded the onset of cardiopulmonary complications during hospitalization. The patient slowly recovered with progressive resolution of symptoms and was discharged after 16 days.

DISCUSSION

Tetanus is an infection that results from wound contamination by the spore-forming bacterium *Clostridium Tetani*. Tetanus is an acute and often fatal disease that is characterized by generalized increased rigidity and convulsive spasms of the musculature. The spores of *Clostridium Tetani*, which represents the quiescent form of the organism, are commonly found in the soil and in animal faeces. The spores enter the body through skin wounds, and, after germination under low-oxygen conditions, excrete a potent toxin, tetanospasmin, into the blood stream. (12)

Tetanospasmin is the neurotoxin responsible for the characteristic clinical features of the infection. After entering the presynaptic terminal in the neuromuscular plate of motor neurons, it inhibits the neurotransmitters release and paralyzes the muscular fibres. The toxin is also capable of retrograde axonal transport to the neurons of the central nervous system. The central neurons, likewise, become incapable of neurotransmitter release. The major central inhibitory neuronal cells, the glycinergic and gamma aminobutyric acid-ergic neurons

(GABAergic neurons), are particularly sensitive to tetanospasmin. Intoxication of these cells leads to the failure of inhibition of motor reflex response to sensory stimulation. The failure of central inhibition results in generalized contraction of the agonist and antagonistic muscles, which characterize a tetanic spasm (13).

Tetanus is classified into four symptomatic types: neonatal, generalized, which represents the most frequent form (more than 80%), local and cephalic (14). In the majority of cases, neonatal tetanus affects the newborn because of contamination of the umbilical stump in an unimmunised mother. Local tetanus is characterized by muscle rigidity near the site of injury or spore inoculation. Cephalic tetanus is a rare type of localized tetanus characterized by the involvement of the cranial nerves in the facial area. Either local or cephalic tetanus may progress to the generalized form (13).

The muscular rigidity usually first involves the masticatory muscles with subsequent lock-jaw, then the disease usually diffuses with a descending pattern with systemic muscular involvement (14).

There are no specific laboratory alterations of tetanus and diagnosis is exclusively based on clinical findings. In the majority of cases (50% to 75%) trismus, due to spasm of the masseter muscle, represents the first sign, followed by spasm of the facial muscles, dysphagia, pain and stiffness of neck musculature (13, 15).

The spatula test could be useful in suspected tetanus. The posterior pharyngeal wall is touched

with a spatula and a reflex spasm of masseter muscles occurs (positive test) instead of the normal gag reflex (negative test) (16).

The differential diagnosis of trismus includes temporo-mandibular joint disorders (ankylosis, arthritis, and disk pathology) and extra-articular causes such as odontogenic infections involving the masticator space; non-odontogenic infections (i.e. tonsillitis, parotid abscess, meningitis); trauma inducing mandibular or zygomatic fractures; pharyngeal or parotid tumours; radiotherapy of the head and neck; and other uncommon causes such as lupus erythematosus and drugs (phenothiazine, succinyl choline, tricyclic antidepressant, metaclopramide, halothane) (17).

Clostridium Tetani is isolated from infected wounds in about 30% of cases. Symptomatic infection most frequently affects individuals with low or undetectable serum levels of anti-tetanus antibodies (<0.1 IU) (17). However, the evidence of protective levels of circulating antibodies is not sufficient to exclude the infection; in fact, cases of tetanus have been described in immunized subjects (15, 18).

Potential life-threatening complications of tetanus include pneumonia (30%) due to aspiration and laryngospasm, rhabdomyolysis (13%), upper gastrointestinal bleeding (9%), cardiovascular instability (9%) (transient cardiac arrest, tachy/bradycardia, arrhythmias and hypertension) due to stimulation of the autonomic nervous system, acute renal failure (4%) and secondary wound



Fig. 1. Panoramic radiograph showing diffuse periodontal disease, without specific signs of dental abscess (e.g. periapical abscess or alveolar osteolytic lesions).

infection (4%) (19-21). Mortality usually results from respiratory failure, cardiovascular collapse, or associated autonomic dysfunctions (7).

Treatment usually requires admission to the intensive care unit (ICU) to control the airways, mechanical ventilation support, control of neuromuscular spasm, and maintenance of the circulation with administration of intravenous fluids. Acute respiratory failure can require endotracheal intubation and even tracheostomy, and often results in prolonged ICU stays, with high risk of secondary opportunistic infections (19).

Passive immunotherapy with HTIG helps to remove released tetanospasmin, but cannot eliminate the toxin that is bound within the nervous system. The recommended dose of intramuscular HTIG ranges between 500 IU and 1.000 IU (15, 17, 18, 22).

Debridement of the contaminated wound with removal of devitalized tissue and other foreign bodies is essential to eradicate the source of toxin production. Given that the disease is caused by a toxin, the efficacy of the antibiotic therapy is still unclear. Penicillin, which had been largely used in the past, is not recommended because of its potential synergic effect with tetanospasmin. Metronidazole has shown good efficacy in controlling the progression of the disease. At present, an intravenous dose of 500 mg four times a day is recommended as first-line therapy (15, 18, 23).

Supportive care includes sedation, neuromuscular blockade and management of autonomic instability. Continuous infusion of diazepam or midazolam (which are GABA-agonist benzodiazepines) is also recommended to prevent the development of respiratory and cardiovascular life-threatening complications. Other drugs, such as propofol, vecuronium, pancuronium and baclofen, had been successfully used as antispasmodic agents (17, 20, 24).

Although tetanus has become a rare disease in developed countries, it remains a potential fatal condition without prompt and aggressive management. Elderly people who live in rural settings are especially vulnerable, because of the declining of immunity to tetanus and the high risk of infected injuries. It is therefore necessary for all health care professionals, and especially for oral and maxillofacial surgeons, to be aware of its clinical

presentation; and the diagnosis should be suspected in all cases with a history of contaminated cutaneous lesion associated with signs of muscular spasm.

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