CASE REPORT

Trismus, the first symptom in a challenging diagnosis of Tetanus

Lorenzo Giannini,¹ Alberto Maccari,¹ Valentina Chiesa,² Maria Paola Canevini²

¹Department of Otolaryngology, San Paolo Hospital, Department of Health Sciences, University of Milan, Milan, Italy

²Regional Centre for Epilepsy, San Paolo Hospital, Department of Health Sciences, University of Milan, Milan, Italy

Correspondence to

Professor Alberto Maccari, bmjotorinosanpaolo@gmail.com

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SUMMARY

Tetanus is a severe, life-threatening infectious disease present worldwide. The incidence of this disease is very low in developed countries, and practitioners are unfamiliar with its symptoms and signs, resulting in late diagnosis and low recovery rate. Furthermore, main symptoms, such as trismus, are often associated with several confounding factors: these may lead the physician to send patients towards an incorrect diagnostic management and the calling on of wrong specialists. This case focuses on the importance of considering tetanus in the differential diagnosis of trismus associated with systemic symptoms, and discusses the clinical implications of an initial wrong diagnostic pathway.

BACKGROUND

Despite the availability of effective immunisation, tetanus remains a leading cause of morbidity and mortality worldwide—the global incidence of tetanus is estimated to be 1 million cases per year.¹ This dangerous disease is rarely encountered in developed countries; however, many cases are still reported, with Italy more frequently stricken than other European states. Indeed, from 2001 to 2011, 594 Italian cases of infection were notified and 169 deaths were reported.²

Tetanus is a severe toxin-mediated infection caused by bacterium *Clostridium tetani*, a non-invasive Gram-positive anaerobic and spore-forming bacterium. Tetanus spores remain localised in the infection site, often a contaminated wound.^{1–3} Once produced, a tetanic toxin called tetanospasmin spreads in the nervous tissue and reaches the inhibitory interneurone of the motor system; it prevents release of γ -aminobutyric acid, thus inducing blockage of inhibitory pathways and muscular spasms.³

Generalised tetanus is the most common form: earlier symptoms usually occur within 2 week after infection, and they include asthenia, trismus, dysphagia and descending muscle rigidity.¹ Usually, trismus is the first symptom in most cases of tetanus (50–75%). However, due to the low incidence of the disease, the physician facing this clinical sign may find it difficult to formulate a correct diagnosis.⁴ Several confounding symptoms and anamnestic factors may delay or hinder a prompt diagnosis. In fact, many conditions are associated with reduced opening of the jaw, or locked-jaw; among these, tetanus is one of the most threatening to the patient's life.⁵

Still, today, diagnosis of tetanus is mainly based on the correct interpretation of symptoms and signs, and the spatula test represents a simple test to diagnose it, with a sensitivity of 94% and specificity of 100%;¹ the posterior pharyngeal wall is touched with a spatula and a reflex spasm of masseters occurs (positive test) instead of the normal gag reflex (negative test).⁴

Once suspected, human tetanus immunoglobulins should be given as soon as possible and intravenous metronidazole should be administered for 7–14 days. Treatment in an intensive care unit (ICU) should always be established, in order to prevent asphyxia and to control the ventilation.¹

CASE PRESENTATION

A 73-year-old woman, otherwise in good health, presented to our clinic with trismus, sore throat, difficulty swallowing solid foods, and cervical and floor of the mouth haematoma. She had a history of paroxysmal atrial fibrillation, treated with oral anticoagulation and arterial hypertension. Her oncological history included a glottic cancer treated 3 years prior with surgery and radiotherapy.

INVESTIGATIONS

Head and neck physical examination revealed moderate trismus (mouth opening of 25 mm) in a total edentulous mouth, and a midline cervical and oral floor haematoma, mildly dislodging the tongue upwards (figure 1). Temporal artery palpation did not evoke pain and found neither thickening nor nodularities. Upper airway endoscopy with flexible endoscope did not show significant airway

Figure 1 Oral examination in moderate trismus: the floor of the mouth is cumbered by a central haematoma, surrounded by hyperaemia; complete mandibular edentulism is present.

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obstruction, but reported mild oedema and hyperaemia of vocal cords, and of the base of the tongue. Complete blood count and blood clotting tests were within normal range. The patient was ordered a maxillofacial and neck CT scan with contrast, which showed a mass in the oral floor without contrast enhancement (figure 2).

DIFFERENTIAL DIAGNOSIS

The woman was hospitalised in order to administer intravenous antibiotic (ceftriaxone 2 g/day and metronidazole 500 mg 3 times/day) and steroid (betamethasone 4 mg 2 times/day) therapy.

Symptoms such as asthenia, malaise and trismus kept worsening despite the therapy, and the cervical/oral haematoma persisted. A few days after admission, the patient presented onset of dyspnoea, requiring assisted ventilation in the ICU, and a tracheostomy was performed. During the procedure, surgeons explored the floor of the mouth, taking a specimen from the central zone of the haematoma, which had a solid, tumour-like appearance. The histological examination of the specimen was inconclusive, finding only traces of inflammatory cell infiltration.

Subsequently, the patient was moved back to the ear, nose and throat (ENT) department; however, no improvement of general conditions was achieved. She showed progressive difficulty in walking, loss of autonomy in daily activities and addiction to oxygen therapy. Neurological impairment required a brain, brainstem, maxillofacial and rachis MRI, which showed no anomalies.

After a psychiatric examination did not reveal anything unusual, a second neurologist was called in. Finally, 19 days after hospitalisation, a tetanus infection was suspected, and confirmed by the spatula test. A more detailed case history revealed that the woman practised gardening, with no tetanus immunisation. No wounds were found during a second physical examination.

TREATMENT

The patient was started on a therapy with intravenous human tetanus immune globulin and continued with intravenous metronidazole.

Figure 2 CT scan, sagittal plane: evidence of an oral, hypodense mass, situated deeply in the body of the tongue.

OUTCOME AND FOLLOW-UP

An improvement of general conditions was rapidly noticed and the patient was moved to the rehabilitation ward 1 week later, where specific rehabilitation exercises for muscle force and respiratory force recovery were performed. After 3 weeks, the tracheostomy could be closed and the patient was discharged 3 months after hospitalisation, with no sequelae of any kind.

DISCUSSION

The presented case shows the difficulty in formulating a correct diagnosis of tetanus despite the presence of many typical symptoms. Trismus is probably the main and earliest sign of the disease, although limited opening of the jaw is a common symptom in general clinical practice. It is often due to trauma or local infection, but can also be the result of psychogenous muscular tension. Less commonly, it is a temporomandibular joint disease, which can be intra-articular, intracapsular or pericapsular (table 1).⁵

In an oncological setting, trismus may also represent a direct effect of the neoplasm on the masticatory muscles as well a consequence of therapy, such as radiotherapy-induced fibrosis.⁶

Moreover, giant cell arteritis with jaw claudication should be suspected in elderly women, and the signs of temporal artery flogosis have to be investigated. The negativity at the temporal artery examination in our patient led us to exclude this hypothesis.

In this case, the patient showed all the features of generalised tetanus since the occurrence of the first symptoms, but

Table 1 Trismus causes (modified from Scully⁵)

Acute trismus	Subacute trismus	Chronic trismus
Infection	Tumour infiltration of muscles or joint	Soft tissue scar formation or TMJ damage after surgery, trauma
Pericoronitis	Chronic infection	
Odontogenic infection	TMJ disease and pain-dysfunction syndrome	Radiation or burns
TMJ or bone infection		Submucous fibrosis
Tonsillar or pharyngeal infection		Scleroderma
Parotitis		Rheumatoid arthritis
Otitis		TMJ disease and pain-dysfunction syndrome
Tetanus		TMJ ankylosis
Trauma		Masticatory muscle disorders
Jaws		Suprabelbar palsy
Facial soft tissues		Giant cell arteritis with jaw claudication
Postoperative		
Third molar teeth removal		
Other jaw and oral surgery		
Associated with haematoma		
Local anaesthetic injection trauma		
Others		
Drug related		
Malignant hyperthermia		
TMJ disease		
TMJ, temporomandibular joint.		

physical examination and medical history led us to focus on the local manifestation. In fact, the patient underwent chemoradiation for glottis cancer, which could have explained the limited jaw opening. In addition, simultaneous occurrence of a floor of the mouth haematoma diverted attention from the overall issue and suggested an oral tumour recurrence. Only one case of spontaneous intraoral mass associated with trismus in tetanus has been reported in the literature; it was later found to be due to forced removal of the patient's denture the day before hospitalisation.⁷ In this case we cannot exclude this eventuality, as the patient had a denture and took oral anti-coagulant therapy for her atrial fibrillation. However, the oral mass may have occurred more likely because of muscle rupture after prolonged spasm of the musculature on the floor of the mouth.⁸

The reason why we did not consider the hypothesis of tetanus earlier may lie in the act of referring the patient to the ENT department, instead of a neurology ward. This is actually common practice, since patients presenting with trismus, masseter spasms, nuchal rigidity and dysphagia tend to consult otolaryngologists, dentists and maxillofacial or orthopaedic surgeons first.⁹ On the other hand, people with convulsions, difficulty in walking and unconsciousness are promptly sent to the emergency room, internal medicine, neurosurgery or neurology department—this means that patients with clear evidence of generalised tetanus do not come to the ENT department's attention.⁹ Therefore, interdisciplinary communication between specialists is pivotal; also important is the training of otolaryngologists and maxillofacial surgeons on recognition of tetanus.⁹

Our diagnostic pathway may have taken a wrong direction due to the nuanced clinical presentation of the early symptoms. Furthermore, the consultation by internal clinicians, neurologists and psychiatrists led us to the false belief that no neurological/psychiatric disease was present. Since they were visiting a ENT patient, their assessments were possibly influenced by our prior considerations. This suggests that proper addressing of the patient to the department of destination from the occurrence of the first symptoms is crucial for applying a proper diagnostic and therapeutic flow-chart.

Learning points

- In case of trismus associated with systemic symptomatology and other apparently unconnected clinical signs, a life-threatening pathology such as tetanus must be promptly suspected in order to avoid fatal complications.
- Tetanus incidence is tenfold higher in Italy than in other countries such as USA, Australia, Germany and the Netherlands; hence, even if the low incidence in the occidental world makes *Clostridium tetani* very unfamiliar for clinicians, Italian practitioners will face this disease more frequently than clinicians in other countries.
- Remaining vigilant on the issue of tetanus is crucial in order to filter pointless hypotheses and establish all the necessary measures for early and lifesaving treatment.

Contributors AM performed surgery. LG wrote the article. MPC and VC played a crucial role in the final diagnosis.

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