Case Report

Picking The Odd One Out: Cephalic Tetanus – A Case Report From Rural Pakistan

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Abstract

Tetanus is an infectious disease with a high risk of mortality. It is caused by an exotoxin, tetnospasmin, produced by Clostridium tetani, an anaerobic gram positive bacillus. The risk of disease is high in the general population due to the abundance of spores in the environment. There are four clinical types of tetanus, generalized, localized, neonatal and cephalic. Cephalic tetanus is one of the rarest clinical form that presents with trismus and cranial nerve involvement. Cephalic tetanus can be difficult to diagnose since it doesn't often manifest with the typical tetanus symptoms like muscle spasms and stiffness. This case is of a 55 year old male who presented to a tertiary care hospital with symptoms consistent with Bell's palsy for 5 days that progressed to development of trismus and neck muscle stiffness with difficulty of breathing that lead to his referral from rural healthcare setup. The case highlights the importance of detailed history and examination with high grade suspicion to make a clinical diagnosis of cephalic tetanus in a country with epidemiological evidence of disease.

Keywords: Cephalic tetanus, Bell's palsy, Infectious disease

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Introduction

Tetanus is a disease that causes tonic muscular spasms as it advances, caused by Tetanospasmin, an exotoxin from the anaerobic Clostridium tetani bacteria. There are four clinical types of tetanus, generalized, localized, neonatal and cephalic. Cephalic tetanus is one of the rarest clinical form, it impacts the cranial nerves and accounts for 0.9-3% of cases overall.¹ Cephalic tetanus can be difficult to diagnose since it doesn't often manifest with the typical tetanus symptoms like muscle spasms and stiffness. The combination of trismus with paralysis of one or more cranial nerves is known as cephalic tetanus and the facial nerve is mostly involved.² There is a lack of case reports of cephalic tetanus therefore, it is a clinical topic to further explore.

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Case Presentation

A non-diabetic, normotensive, nonsmoker 55 years old male presented to the Emergency Department of Holy Family Hospital from a rural area of Pakistan with the history of deviation of angle of mouth to right side and drooling of saliva for last 5 days. He also developed difficulty in opening mouth leading to inability to eat and chew for last two days. He was being managed on lines of Bell's palsy at the periphery. One day before presenting to tertiary care hospital, he developed stiffness of neck muscles with difficulty in breathing and worsening jaw stiffness. He denied any spasm in other parts of body. There was no history of fits or loss of consciousness. Past history revealed that he had a fall while working in the fields about two weeks ago resulting in small lacerations over his knee and face. He took analgesics and local dressing of the wound but did not receive tetanus toxoid booster dose. History for past vaccination status was also negative for any immunization.

On examination he was anxious and agitated, with the following vitals; heart rate 180/min with regular rhythm, blood pressure 120/70 mmHg, and respiratory rate 16/min and oxygen saturation 92% at room air. Mouth

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opening was barely one finger across, angle of mouth was deviated to the right side, there was failure to close left eye with loss of nasolabial fold on left side. Facial palsy of lower motor neuron type with House-Brackmann Grade V was observed. He had stiffness of neck muscles (Figure 1 a-c) and difficulty breathing. The rest of the systemic examination was unremarkable. A diffe-



rential diagnosis of cephalic tetanus was made on clinical grounds with conditions like brain stem stroke and other causes of Bell's palsy that were excluded.

Investigations including baselines blood tests (table $\underline{1}$) and radiological imaging came out to be normal. Renal and liver function tests were also normal. CT scan brain was unremarkable. Nerve conduction study wasn't nece-

Test Name	Patient's Value	Reference Value
Hemoglobin (Hb)	16.5g/dl	14-17 g/dl [<u>3]</u>
Total Leucocyte Count (TLC)	10100/µ1	4500-11000/µ1 [<u>3</u>]
Platelet Count	135,000/µl	150,000-350,000/µ1 [3]
Serum Sodium	144mEq/1	135-146mEq/L [3]
Serum Potassium	4.3mEq/1	3.5-5.0mEq/L [3]
Serum Calcium	9.3mg/dl	9-10.5 mg/dl [<u>3]</u>

ssary because of the peripheral character of the facial paralysis, its short duration of evolution, and the absence of traumatic cause to give an idea of the possible interest of surgical exploration.

Patient was initiated on the treatment for cephalic tetanus; injection tetanus toxoid and tetanus immunoglobulin along with intravenous antibiotics metronidazole and ceftriaxone. Intravenous midazolam was also added. Consent was taken for tracheostomy, but the breathing difficulty and neck muscle stiffness improved with treatment, so it was deferred. Patient was managed in Medical ICU for a week and showed marked improvement in the symptoms and discharged in stable condition.

Discussion

Tetanus is an infectious disease with a high risk of mortality. It is caused by the spores of Clostridium tetani,

an anaerobic gram positive bacillus. The risk of disease is high in the general population due to the abundance of spores in the environment, particularly soil, ash, animal excreta and rusty tools. The disease burden has been fairly reduced from the world as a result of effective immunization strategies, however, it is an important public health concern in developing countries due to lack of vaccination.⁴ Global incidence has decreased from an estimated 615,000 cases globally in 1990 to about 74,000 in 2019.⁵ In the year 2020, tetanus accounted for 0.52% deaths in Pakistan. The age adjusted death rate of 2.15 per 100,000 of population ranks Pakistan 7th in the world.⁶

Cephalic tetanus is a rare form of tetanus that manifests as trismus and cranial nerve involvement. Facial nerve is the most commonly affected cranial nerve. Cephalic tetanus accounts for 1 to 3% of the total number of reported cases of tetanus and has a mortality of 15 to 30%.⁷ The mode of transmission is through spore contamination of a craniofacial injury or secondary to otitis media.⁸ Cases of cephalic tetanus arising from dental origin have been noted." Rare incidences have been reported where any injury could not been identified making the diagnosis fairly challenging.^{10,11} In our case, the portal of entry was through injury on the face as a result of fall in the field which was brought into knowledge through history. Tetanus is a clinical diagnosis which is supported by the epidemiological evidence of disease in the community, therefore thorough history and examination with high grade suspicion should be relied on for initiating the treatment. There is no diagnostic investigation for tetanus, however, serum immunoglobulin levels prior to the administration of anti-tetanus immunoglobulin can be of some help, although the disease has been described in the presence of antibody levels that are considered to be protective.¹⁰ Tetanus can be misdiagnosed as a number of medical emergencies including electrolyte disturbances, seizures, meningitis and stroke, but these conditions usually lack the characteristic features of the disease. However, these may cause a delay in diagnosis that may lead to the progress of the disease to generalized form. In our case, the patient was being managed as a case of Bell's palsy before he developed lock jaw and difficulty in breathing. The differentials were ruled out by a normal brain imaging and electrolyte studies and the presence of trismus and neck muscle stiffness suggesting otherwise. Nerve conduction study was not required due to a peripheral lesion of short course of disease with no surgical cause behind it.²

Cases of cephalic tetanus have been reported with only facial nerve involvement of peripheral type. It may also precede the development of trismus like we observed in our case.^{2,12} Since cephalic tetanus is a clinical diagnosis, this presentation is a reason in diagnostic delay, which in our patient was observed to a limited extent owing to the timely referral of the patient to a tertiary care facility.

The clinical diagnosis is followed by immediate treatment with tetanus toxoid, anti-tetanus immunoglobulin, antibiotics and symptomatic management using sedatives, oxygen supplementation and in severe cases, tracheostomy. In the case presented, tracheostomy, although planned, was deferred due to good clinical response of the patient to active and passive immunization and antibiotics.

Conclusions

Tetanus is a pure clinical diagnosis, and in cases of generalized tetanus, diagnosing the case may not be challenging for a physician working in Pakistan as it is common due to poor vaccination status. On the other hand, as the presentation of a case of localized or cephalic tetanus might vary, making it challenging to diagnose it early therefore reporting such cases hold great significance to make physicians aware about it for the best interest and safety of the patient.

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