IDCases 24 (2021) e01147

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

Progressive localized tetanus in patient with inadequate human tetanus immunoglobulin therapy

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ARTICLE INFO

Article history:
Received 21 April 2021
Received in revised form 30 April 2021
Accepted 30 April 2021

Keywords:
Localized tetanus
Localised tetanus
Progressive
Evolve
Generalization

ABSTRACT

Localized tetanus is a rare form of tetanus and generally has a better prognosis, but it could become fatal when it progresses into generalized tetanus. Treatment is challenging as there are limited data about clinical management and factors preventing localized tetanus progression. We present a case of localized tetanus in left lower limb of 8 year-old boy which progressed into generalized tetanus.
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Introduction

Tetanus is a vaccine-preventable disease which still remains a public health problem in many parts of the world, especially in low-income countries or districts, where immunization coverage is low. Localized tetanus is a rare form of tetanus which affect groups of muscles [1,2]. Although localized tetanus generally milder, it could become fatal when it progresses into generalized tetanus. Literature describing generalization of localized tetanus is quite rare to be found [3,4]. In this case report, we present a case of localized tetanus which developed into generalized disease and our management in a limited resources hospital.

Case presentation

An 8-year-old boy from a rural area in Borneo presented with painful muscle spasm localized to his left leg and feet which developed 12 h before. The patient had a history of a puncture injury from a rusty nail in his left heel and did not get any treatment about 10 days before the symptoms. The patient had a history of uncompleted tetanus immunization. His nutritional status are within normal range.

Vital signs showed slightly tachycardia and tachypnea with Wong-Baker Face pain scale at 9–10 during spasm. There were no features of bowel/bladder involvement, loss of consciousness and sensory, signs of generalized tetanus such as trismus, opisthotonus, difficulty in breathing or swallowing. His left lower limb was being held in 90° knee flexion with fully plantar-flexion at the ankle. No movement was possible at knee and ankle joint. The hip was able to move with a strictly limited range of movements. Muscle tone were markedly increased, especially the posterior compartment muscles of the leg and hamstring muscles. A single spasm could persist more than 5 min with relaxation appeared to be less than 30 s, fully relaxation was never reached. Any touch and movement below hip exaggerated the spasm. There was a healed puncture wound on left heel with no signs of inflammation and infection. The other limb was normal.

Laboratory blood investigations showed leukocytosis (12.360/μL) and thrombocytosis (562.000/μL), with normal blood sugar level and serum electrolytes. Any further lab investigations could not be done due to limited facilities in our hospital. Radiographic imaging on left lower limb showed no bone, joint or soft tissue abnormalities.

Based on history taking, physical examinations and laboratory findings, diagnosis of localized tetanus was made and human tetanus immunoglobulins (HTIG) were given immediately. Patient could only receive 250 IU HTIG during the first injection due to a limited amount of the preparation in hospital. HTIG dose during first injection was less than any recommendations and considered

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http://dx.doi.org/10.1016/j.idcr.2021.e01147
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inadequate. Adequate 3000 IU HTIG therapy as American Academy of Pediatrics recommendation was achieved 8 h after the first HTIG injection [5].

Wound debridement and first antibacterial administration were performed soon after first 250 IU HTIG injection. Ceftriaxone (100 mg/kg/day) and metronidazole (30 mg/kg/day) were given as antibacterial coverage for 14 days. The limb spasms were treated with intravenous diazepam (0.15 mg/kg/dose) and repeated every 4 h. The patient was treated in the isolation room with minimal lighting and noise.

After antibacterial administration, the spasm showed progression to the right lower limb and ascended to trunk and neck, showing sign of opisthotonus until the first 24 h. Since the spasm tended to appear only after the diazepam effect subsides, diazepam administrations were repeated more often to every 2 h with close monitoring on signs of respiratory depression and hemodynamics instability.

The spasms showed improvement on treatment. The diazepam was repeated less often and switched into oral diazepam after 2 weeks. The patient was observed and rehabilitated for the next 2 weeks until he could stand and walk. He was able to stand and walk at the time of discharge, even though the range of motion of his left knee and ankle were still limited (Fig. 1).

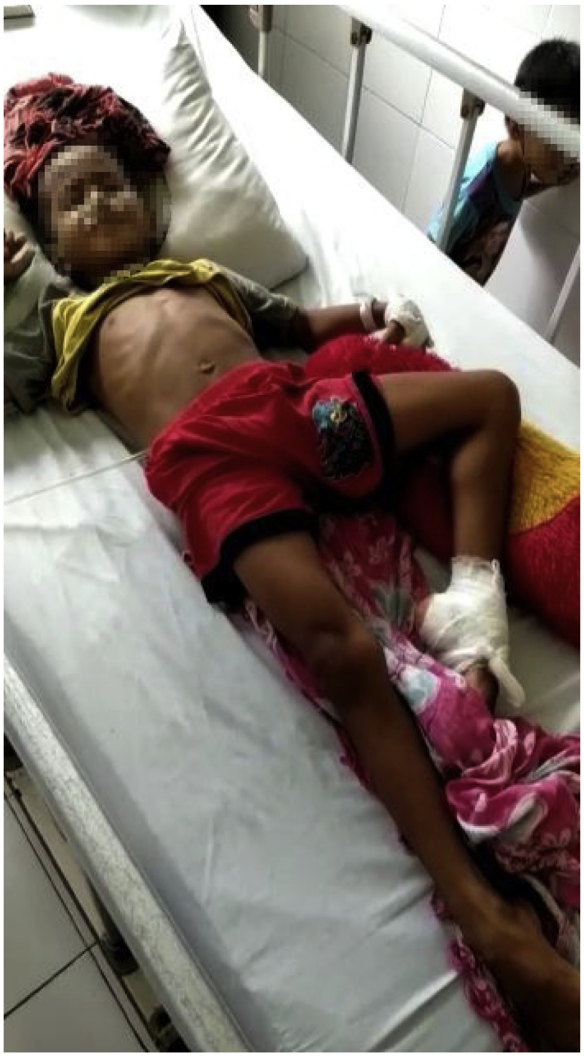

Fig. 1. Clinical presentations of localized tetanus on left lower limb with sign of opisthotonus after 24 h of treatment.

Discussion

Tetanus is a serious and potentially life-threatening infectious disease caused by tetanus neurotoxin (TeNT) produced by *Clostridium tetani* [1,6]. TeNT blocks the release of neurotransmitter on inhibitory neurons causing severe unregulated muscle contraction and spasm [7].

Localized tetanus is a rare form of tetanus which affect groups of muscle near to the injury and generally milder [1,2]. It is reported about 2 %–13 % of tetanus cases [3,7–9], but the actual prevalence of localized tetanus remains unknown since it is only reported in a few reports. In several cases, localized tetanus can be fatal when it progressed into generalized tetanus [1]. Retrospective studies in Abidjan found that patients with localized tetanus have a 27 % risk of developing secondary generalization, 16 % mortality rate and 11 % are have persistent sequelae [3].

The diagnosis of localized tetanus is entirely clinical [1]. Bacteriologic confirmation is not needed since *C. tetani* is difficult to culture and any wound can be contaminated by *C. tetani* without developing tetanus [7]. Appropriate tetanus immunization and protective anti-tetanus antibody titer might be helpful, but do not exclude the possibility of developing tetanus. Evolving localized tetanus has occurred despite prior immunization and protective antibody titer [4]. In our case, diagnosis was made clinically based on the presentation of muscle spasm localized to left feet and leg, without any loss of consciousness and signs of generalized tetanus, supported with patient history of uncompleted tetanus immunization and untreated punctured injury in his left heel prior to symptoms.

The chance of patient developing localized tetanus and its progression are known related to patient factors (immune status, nutritional status), vaccine factors (vaccine quality, improper interval dosing of vaccines) or improper wound management [4]. In our case, patient history of uncompleted tetanus immunization may play a role in the manifestations of localized tetanus.

TeNT is presumably released when *C. tetani* cells break open [10] and can't be neutralized once bound to nerves [1]. This has implications for the treatment of tetanus. HTIG therapy prior to antibacterial administration may play critical role in neutralizing excess toxin released once the antibacterial destroys the bacterial cells. American Academy of Pediatrics recommend giving 3,000–6,000 IU HTIG in a single dose for tetanus therapy, as CDC and WHO recommend giving 500 IU HTIG in a single dose [1,5,11] However, the optimal therapeutic dose of HTIG have not been established [5].

Until now, no literature mentioned about prior HTIG therapy to antibacterial administration and its implications / correlations with progression of localized tetanus. We assumed that there is a possibility that our initial antibiotic therapy before adequate passive immunization due to lack of HTIG preparation might be the cause of progression.

Conclusion

In conclusion, localized tetanus has a better prognosis than generalized tetanus. Localized tetanus could evolve to generalized tetanus, but our understanding about which factors determined progression of localized tetanus are still limited. Adequate HTIG therapy prior to antibacterial might prevent the progression of localized tetanus. However, the optimal therapeutic dose of HTIG and its timing related to antibiotics administration have not been clearly established. Further scientific study and analysis about this topic are needed to provide new insights and guidelines on clinical management of localized tetanus.

Funding

There has been no significant financial support for this publication. Authors would bear the cost of published paper.

Ethical approval

Ethical approval was obtained from ethical committee at out hospital.

Consent

Informed consent was obtained from the patient for publication of this case report and accompanying image/video.

Author contribution

DL conceptualization, design of study, data collection, analysis and interpretation, literature search, writing manuscript. EMP data analysis and interpretation, writing manuscript. MR providing patient, revision of the manuscript. HL providing patient, added some literature, supported data analysis and interpretation, revision of the manuscript. All authors read and approved the final manuscript.

Declaration of Competing Interest

We know of no conflicts of interest associated with this publication.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.idcr.2021.e01147.

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