

**Case Report**

Cutaneous Diphtheria Necrotizing Fasciitis of the Upper Limb. A Rare Case Report.

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Background: Diphtheria is a respiratory and cutaneous disease caused by toxicogenic *Corynebacterium* which produce diphtheria toxin capable of affecting mainly mucus membranes and to a certain extent subcutaneous tissues. Only a few cases of necrotizing fasciitis of the upper limb have been reported in the literature.

Case Report: An 18-year-old previously vaccinated gentleman presented to our Emergency Unit with a stab on the left forearm with a knife. The wound was a 4cm penetrating incised wound on the radial aspect of the proximal forearm. There were no signs of contamination and he was routinely thoroughly cleaned and sutured and discharged the same day. He presented a week later with necrotising fasciitis of the forearm extending to the hand and arm. Standard radical debridements were done and the microbiology from the specimens isolated *Corynebacterium diphtheriae*.

Discussion: This case highlights a rare case of Diphtheria ulcerans presenting as Necrotising Fasciitis on a patient who was previously vaccinated (for Diphtheria as a child). In most third world countries, the Vaccine is mandatory in the form of DPT (Diphtheria, Pertussis and Tetanus) as a complete vaccine series. A literature search revealed only one case of upper limb diphtheria necrotising fasciitis, and some other reports mentioned sporadic resurgence of diphtheria infections in 1st world countries where the vaccination is not mandatory. Some with a travel history to third world countries...

Conclusion: Necrotizing fasciitis caused by diphtheria is extremely rare and further interrogation of the relationship between vaccination and the resurgence of diphtheria infections in mostly 1st world countries must be undertaken.

Keywords: Necrotizing; Fasciitis; Diphtheria; Upper limb.**Introduction**

Diphtheria is a respiratory and cutaneous disease caused by mainly two species: *Corynebacterium diphtheriae* and *Corynebacterium ulcerans*, and to a lesser extent by *Corynebacterium pseudotuberculosis* [1]. These are toxicogenic *Corynebacterium* which produce diphtheria toxin capable of affecting mucus membranes and subcutaneous tissues.

In most African countries where Diphtheria vaccinations are compulsory, only isolated cases have been described over the

years. In South Africa, the last significant outbreak was in 2015 when 15 cases of respiratory diphtheria were discovered [2], 75% of which were in not vaccinated individuals. In October 2023, the National Institute for Communicable Diseases (NICD) of South Africa issued an alert of 12 laboratory-confirmed cases of toxigenic *Corynebacterium diphtheriae* in the country [3] and the following month, the Ministry of health announced the identification of 8 more cases in a new outbreak in a prison [4]. Our case was an isolated case identified in a tertiary facility the same year.

Cutaneous manifestation of the disease is thought to be associated with zoonotic transmission [5-9] or travel to endemic destinations

[10] by the unvaccinated. A report from Canada [11] associated the cutaneous diphtheria with low socio-economic status.

Cutaneous diphtheria is typically ulcerative not cellulitic and it has been reported to begin with an innocuous pustule or vesicle which eventually develops into a well-formed punched out superficial ulcer. They typically occur in the hands, the feet, and legs. The ulcer is initially painful, becoming asymptomatic with time and it is normally self-limiting [12]. Systemic manifestations, especially of the toxicogenic species can also occur in about 15% of patients including but not limited to fevers, rigors, tachycardia with myocarditis, and peripheral neuropathies [13]. Cases of necrotizing fasciitis due to either *Corynebacterium ulcerans* or *Corynebacterium diphtheria* are rare. A case was reported in Germany in 2012. [14].

Case Report

We present a case of Cutaneous *Corynebacterium diphtheria* presenting as necrotizing fasciitis of the hand and forearm isolated in a tertiary institution in South Africa. An 18-year-old gentleman presented to the Surgical Emergency Unit (SEU) with a history of a stab on the left forearm with a knife. He had sustained a deep laceration of the left forearm with muscles exposed and he was unable to mobilize the left arm. Examination confirmed a 4cm penetrating incised wound on the radial aspect of the proximal forearm which was not actively bleeding, and he was neurologically intact. There were no signs of contamination or sepsis at that stage and the radiological investigations were not remarkable.

He was thoroughly cleaned and sutured in the emergency room and then discharged home with instructions to follow-up at a local clinic. No antibiotics were prescribed at that stage. The patient presented a week later with a history of progressive left arm swelling and pus draining from the suture site. He also complained of an inability to mobilize the left arm and he was in severe pain. On examination, the left arm was markedly swollen from axilla to hand, with pus oozing from the suture site and the arm was warm and tender. X-rays were not remarkable, and Ultrasound showed cobble stone appearance consistent with cellulitis, with no deep tissue collections.

The LRINEC score [15] calculation was worked out at 9 suggesting a 75% chance of the diagnosis of Necrotizing fasciitis. He was adequately resuscitated and started on an empiric course of antibiotics (Clindamycin, Metronidazole and Augmentin) while he was being prepared for radical debridement. He was debrided on the same day with a long lazy S fasciotomy of the forearm.

All necrotic tissues were debrided, and specimens were sent for Microscopy Culture and Sensitivity (MC& S) and cultured *Corynebacterium diphtheria* infection which was non-toxin producing. Our institution infection control unit was notified as it is a notifiable disease, and they suggested not to initiate the vaccine as the bacteria was non-toxin producing. They further advised to complete the course of the empiric regimen he was already on. We subsequently took him for a relook in 48 hrs in theatre and the wound was found to be clean with no extension of the fasciitis. After two more debridements, the sepsis was deemed cleared and a planned cover with a split thickness skin graft was done successfully. Infection control unit of our hospital then suggested expectant treatment with a shot of vaccine, and this was duly administered. There were no other foci of infection anywhere in the body and he never exhibited any of the classic signs and symptoms of Diphtheria (throat). Further questioning could not reveal whether he indeed had a vaccine but for the fact that the vaccine is compulsory for all infants born in the public facilities in South Africa, we concluded that he did indeed have the vaccine in his childhood. At our last review, 6 weeks later, the skin graft had fully incorporated, and he had regained full function of the limb. He has since been discharged and followed up by National Institution of Communicable Diseases (NICD).

Discussion

This case highlights a rare case of Diphtheria ulcerans presenting as Necrotising Fasciitis at our institution on a patient who was previously vaccinated (for Diphtheria as a child). In most third world countries like South Africa, the Vaccine is mandatory in the form of DPT (Diphtheria, Pertussis and Tetanus) as a complete vaccine series. CDC reports that a combination vaccine (DT) has a clinical efficacy of 100% for tetanus and 97% for diphtheria. A complete series is three doses for people 7 years or older and four doses for children younger than 7 [16].

We did a comprehensive literature search and found only one report of *Corynebacterium diphtheria* being the causative organism in Necrotising fasciitis of the upper limb. What really raises the question is whether there is a possibility of mutations occurring in the bacteria altering their virulence because of inadequate vaccinations, especially in most first world countries where they are not mandatory. As mentioned earlier, most of the cases reported in the literature are of patients who travelled from the first world countries to endemic areas. Further interrogation of this finding would be interesting (Figures 1-4).



Figure 1: Close up at presentation.



Figure 2: Dorsum Post Vaccum.



Figure 3: Post Vaccum.



Figure 4: The arm at presentation.

Acknowledgments: The department of Orthopaedics at CHBAH and all the interns who helped with the Necrotizing fasciitis data collection.

Ethics: The authors declare that this submission is in accordance with the principles laid down by the Responsible Research Publication Position Statements as developed at the 2nd World Conference on Research Integrity in Singapore, 2010. Consent was obtained from patients for the use of clinical photographs/ and these images were adequately anonymised. The research ethical approval was obtained from the Human research ethical committee (HREC (MEDICAL)) University of the Witwatersrand, Johannesburg, South Africa.

Conflict of interest: The authors confirm that there were no conflicts of interest

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