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CASE REPORT

Necrotizing fasciitis induced by *Vibrio vulnificus* in patients without marine contact in Hong Kong



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TRANSLATION

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Abstract Necrotizing fasciitis caused by Vibrio species is a life-threatening soft tissue infection with rapid progression and high mortality. The classic history of Vibrio species—induced necrotizing fasciitis is the infection of wounds by direct invasion or contact with contaminated seawater or raw seafood, especially in immunocompromised patients.

We present two cases of *Vibrio vulnificus* necrotizing fasciitis in the upper limb without any wounds or seawater contact and with good past medical history. Both underwent timely surgical debridement and resulted with good functional outcome. Although rare, as clinicians, we need to have a high index of suspicion for the possibility of *V. vulnificus* necrotizing fasciitis despite no risk factors and give timely and appropriate treatment and, more importantly, patient survival.

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Introduction

Necrotizing fasciitis caused by Vibrio species is a lifethreatening soft tissue infection with rapid progression and high mortality. Immunocompromised patients are also at higher risk of developing this disease. *Vibrio vulnificus* are found in warm coastal waters, typically exceeding 20 °C, which is ideal for the growth of the microorganism [1]. The typical history of Vibrio species—induced necrotizing fasciitis is the infection of wounds by direct invasion or contact with contaminated seawater or raw seafood [2]. The infection can predispose the patient to fatal sepsis and multiorgan failure within 24 h. Early diagnosis and emergency fasciotomy or amputation and antibiotics are,

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therefore, crucial in reducing mortality in patients, which can reach as high as 25-100% [2].

We present two cases of *V. vulnificus* necrotizing fasciitis in the upper limb without any seawater contact and with good past medical history. Both underwent timely surgical debridement and resulted with good functional outcome. Although rare, as clinicians, we need to have a high index of suspicion for the possibility of *V. vulnificus* necrotizing fasciitis despite no risk factors, and give timely and appropriate treatment and, more importantly, patient survival.

Case report 1

A 67-year-old man, retired manual worker, non-smoker, non-drinker with good past health, attended the accident and emergency department in the morning complaining of left elbow swelling and pain after being bitten by an unidentified insect.

On examination, his temperature was 38.5 °C and his blood pressure on the low side, which responded with intravenous fluids. There was swelling and erythema located over the medial aspect of his left elbow. An 8-mm clear blister and associated patchy hemorrhagic pigmentation of the skin was identified.

The white cell count was 8.4/mm³, and C-reactive protein level was 25.1 mg/dL. Laboratory risk indicator for necrotizing fasciitis (LRINEC) score was 4. Bedside exploration and probe test were performed at bedside with no dirty brownish tan—coloured fluid or pus found. He was placed on intravenous antibiotics.

Subsequent clinical examination revealed rapid spread of erythema, with several hemorrhagic blisters formed within 6 h. Blood pressure was persistently low despite intravenous fluids. The clinical diagnosis of necrotizing fasciitis was made, and emergency fasciotomy and debridement were performed.

Intraoperatively, wound exploration revealed foulsmelling discharge of necrotic tissue [3]. The finger test [4,5] also showed the fascial plane opened abnormally easily [6]. Extensive debridement of the forearm fascia and necrotic tissue was performed.

The patient required intensive care unit support for inotropic support. Biopsy of the fascia confirmed necrotizing fasciitis, and culture of the intraoperative dirty brownish-tan—coloured fluid was *V. vulnificus*. Two more serial debridement procedures were performed in the following 48 h. A split-thickness skin graft was performed at 1 week, and the latest follow-up at 2 months showed his elbow had good range of motion from 0 to 130° .

Case report 2

A 55-year-old man, retired mechanic, smoker and social drinker with good past health, attended the accident and emergency department in the afternoon with left forearm swelling and pain. The initial presentation was 6 h before with no preceding causes at home. He had fever of $38.7 \degree C$, blood pressure 88/48 mmHg and pulse 72 bpm. He was drowsy and septic looking. A 4×3 cm hemorrhagic blister was present over the dorsum of the midforearm with

surrounding erythema. The white cell count was 20.7/mm³, and C-reactive protein was 25.7 mg/dL. LRINEC score was 6. The clinical diagnosis was necrotizing fasciitis, and urgent operation was arranged. Extensive debridement was performed with dirty brownish-tan—coloured encountered during the initial incision of the forearm. Postoperatively, the patient was sent to the intensive care unit and required supportive inotropes. Biopsy of the fascia confirmed necrotizing fasciitis, and culture of the intraoperative dirty brownish-tan—coloured fluid was *V. vulnificus*.

Second-look debridement was performed within 48 h. He was discharged from the intensive care unit the next day, and his inflammatory markers normalized 2 days later. He proceeded for full-thickness skin graft subsequently. During his latest follow-up, the wound had healed and his range of motion of his elbow and wrists were full.

Discussion

Necrotizing fasciitis caused by Vibrio species is a lifethreatening soft tissue infection with rapid progression and high mortality. V. vulnificus was first described in 1979 and is a naturally occurring Gram-negative, halophilic bacterium that is an inhabitant of marine environments [1]. Typically, the bacterium is found in warm coastal waters, including Hong Kong. The species produce extracellular enzymes and toxins that cause destruction of local perforating vessels of skin flaps that results in the necrosis of subcutaneous tissue above the deep fascia [1]. The classic history of Vibrio species-induced necrotizing fasciitis is the infection of wounds by direct invasion or contact with contaminated seawater, especially in immunocompromised patients [2]. Necrotizing fasciitis due to systemic V. vulnificus infection and haematogenous spread has also been described [7]. However, most patients have risk factors, and in our two cases, these were not identified. With no definite portal of entry and seawater contact, the pathological process may be different from infection of wounds. Future studies should further identify possible routes of Vibrio infection including insect bites as in our first case and possible haematogenous spread in our second case. More importantly, caution should be taken for any patient presenting with infection and unstable vitals.

Necrotizing fasciitis can predispose the patient to fatal sepsis and multiorgan failure within 24 h. Early diagnosis and emergency fasciotomy or amputation and antibiotics are crucial in reducing mortality in patients [2]. The LRINEC score has been reported to be a useful predictor for the likelihood of necrotizing fasciitis given preliminary laboratory results [8]. The scoring system is derived from six routinely performed investigations and used initially to distinguish early necrotizing fasciitis from other severe soft tissue infections [8]. However, the score has been reported in studies to be inaccurate in necrotizing fasciitis caused by *V. vulnificus* [9].

The primary treatment for necrotizing fasciitis is surgical debridement. Although timely resuscitation and intravenous antibiotics might temporarily stabilize the patient, immediate operation must be initiated if the patient shows any progressive clinical findings or suspicion of necrotizing fasciitis [10]. Because ischaemia and hypoxia compromises the adequate delivery of antibiotics at the infected site, conservative management with antibiotics alone has little role. The use of inotropes is for resuscitation purposes and maintenance of blood pressure, but most importantly, early and aggressive surgical debridement is ultimately needed to treat the infection. Therefore, careful and frequent examination of the patient is essential. Intraoperatively, the extent of debridement is still currently based on surgeon's intraoperative assessment [11], which might not be fully ac-But more importantly, curate [12]. meticulous debridement is essential to save life in cases of necrotizing fasciitis. Extensive debridement should be performed of all tissues that can be easily elevated off the fascia with gentle pressure. Restricted primary debridement has been associated with 7.5 times increased relative risk of death [13].

To our knowledge, there are three documented case reports of necrotizing fasciitis after an insect bite. These insects include a mosquito [14], a tick [15] and an unspecified insect [16] yielding Klebsiella, group A β -haemolytic streptococcus and *Cryptococcus gattii* from microbiology cultures, respectively. It is surprising and unusual that our first patient had contracted *V. vulnificus* necrotizing fasciitis caused by an insect bite as the bacteria is found in marine waters. Furthermore, our patients had good past health, no prior wounds or contact with seawater or seafood. Previous case reports show that patients have risk factors including seawater contact, wounds [17], injury from fish [18], oyster dining [19] and immunocompromised status [20].

Although *V. vulnificus* is a potentially lethal opportunistic pathogen, it remains scientifically unknown what the virulent or fatal dose is for humans [21]. Currently, there is a lack of local epidemiological data in Hong Kong. Given the uncommon encounters, delay in clinical recognition and treatment often leads to poor outcomes [22].

Future studies should show the epidemiological data and pathomechanisms of the mode of disease transmission of *V*. *vulnificus*. Although rare, as clinicians, we need to have a high index of suspicion for the possibility of *V*. *vulnificus* necrotizing fasciitis despite no risk factors and give timely and appropriate treatment, which increase chances of limb preservation, and more importantly, patient survival.

Conflicts of interest

The authors have no conflicts of interest to disclose in relation to this article.

Author contributions

R.M.Y.W., W.-L.T. and P.-C.H. contributed in conception and design of study. R.M.Y.W. and L.T.-C.C. contributed in acquisition of data and drafted the manuscript. R.M.Y.W., W.-L.T., M.C.-K.M. and P.-C.H. contributed in analysis and/ or interpretation data. W.-L.T., M.C.-K.M. and P.-C.H. revised the manuscript critically for important intellectual content. All authors approved the version of manuscript to be published.

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Ethical approval

Ethics approval has been obtained for the study (CRE-2018.192).

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jot.2019.03.004.

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