CASE REPORT

Recurrent *Chromobacterium Violaceum* Infection in a Patient with Chronic Granulomatous Disease

M Sureisen, MBBS, S K Choon, FRCS, C C Tai, FRCS (Orth)

Division of Joint Replacement, Department of Orthopaedic Surgery, University Malaya Medical Centre, 50603 Kuala Lumpur, Malaysia

SUMMARY

We report a rare case of recurrent infection of *Chromobacterium violaceum* in an immunocompromised patient. Despite the high mortality rate associated with this infection as reported in the literature, this patient survived three episodes of recurrent infection. We believe that with high clinical suspicion, prompt treatment and appropriate antimicrobial agents, it is possible for clinicians to treat this infection effectively and reduce the mortality rate.

KEY WORDS:
Chromobacterium violaceum, Immunocompromised, Mortality, Recurrent infection

INTRODUCTION

Infection with *Chromobacterium violaceum* is extremely rarely and there are only few reports worldwide. *C. violaceum* is a gram negative saprophyte isolated from water and soil, commonly in tropical and subtropical areas. The first ever human infected case was mentioned in Malaysia in 1927[1].

C. violaceum infections are associated with rapid dissemination and a high mortality rate due to fulminant sepsis and multi-organ system failure[2]. Although infection with *C. violaceum* has been reported in immunologically competent persons, several reports exist in persons with innate or acquired immune deficiency including chronic granulomatous disease (CGD).

We report a case of a 20-year old patient who suffered from CGD and presented with recurrent episodes of sepsis, multi-organ abscesses and infected non-healing ulcer due to *C. violaceum* over the past eight years. To our knowledge, there have been only two other recurrent infection reported worldwide and both involving immunocompetent patients[3,4]. This appears to be the first ever recurrent infection of *C. violaceum* reported in an immunocompromised patient.

CASE REPORT

In July 1999, a previously healthy 12-year old boy presented with acute abdominal pain associated with fever and diarrhoea. Within a few hours after arrival at the hospital, his condition deteriorated and he developed respiratory distress with hypotension. It was noted on admission that he had a superficial traumatic wound with surrounding cellulitis on his right foot, which was due to a football injury three days earlier in a waterlogged field. He was subsequently intubated and supported with inotropic agents and empirical antibiotics. Within the next 24 hours, he developed multiple organ dysfunction. On the third day of admission, swab culture from his right foot wound grew *C. violaceum* which was sensitive to cotrimoxazole, gentamicin, and ciprofloxacin. His condition improved after he was started on ciprofloxacin. He was later diagnosed to suffer from CGD based on immunological tests. He was discharged home after two weeks of hospitalization.

One year later, he presented with fever and infected wound on his right knee after a minor knee injury. His general practitioner started him on cloxacilin empirically but he did not respond to treatment. He was then admitted to hospital and the culture from his knee wound swabs revealed the growth of *C.violaceum* which was sensitive to cotrimoxazole. Treatment was commenced immediately and he made a full recovery and was discharged five days later. He was advised to complete a further ten days course of oral antibiotics.

In July 2007, he presented with another episode of acute abdominal pain with fever. His left forearm was swollen with a discharging pustular lesion. He could not recall any injury to his forearm. The abdominal examination was normal with no organomegaly. However, he was noted to have enlarged axillary lymph nodes. His white blood cell count was raised (12.9 x 10^9/L) but with normal differential count. Both C-reactive protein (26.8mg/dL) and erythrocyte sedimentation rate were elevated (> 140 mm/hr). The liver function test showed slight elevation of aspartate aminotransferase (43 IU/L) and gamma-glutamyl transpeptidase (100 IU/L). The computed tomography of his abdomen and thorax showed multiple ring enhancing lesions in segment II, V, VI and VIII of the liver and his spleen, and fibrotic changes in both lung apices and lung bases (Figure 1). He underwent urgent incision and drainage of his left arm lesion. The microscopy and culture of pus revealed *C. violaceum* sensitive to ciprofloxacin and cotrimaxole. He responded immediately to treatment and he made an uneventful recovery. He was discharged on the seventh day with prescription of oral antibiotics for another week.

DISCUSSION

Although infection with bacterium *C. violaceum* is rarely encountered, the mortality rate associated with this infection is extremely high. In the United States, 73% of the cases

This article was accepted: 2 October 2008
Corresponding Author: Tai Cheh Chin, Department of Orthopaedic Surgery, University of Malaya, 50603 Kuala Lumpur, Malaysia
Email: tai.chehchin@gamil.com

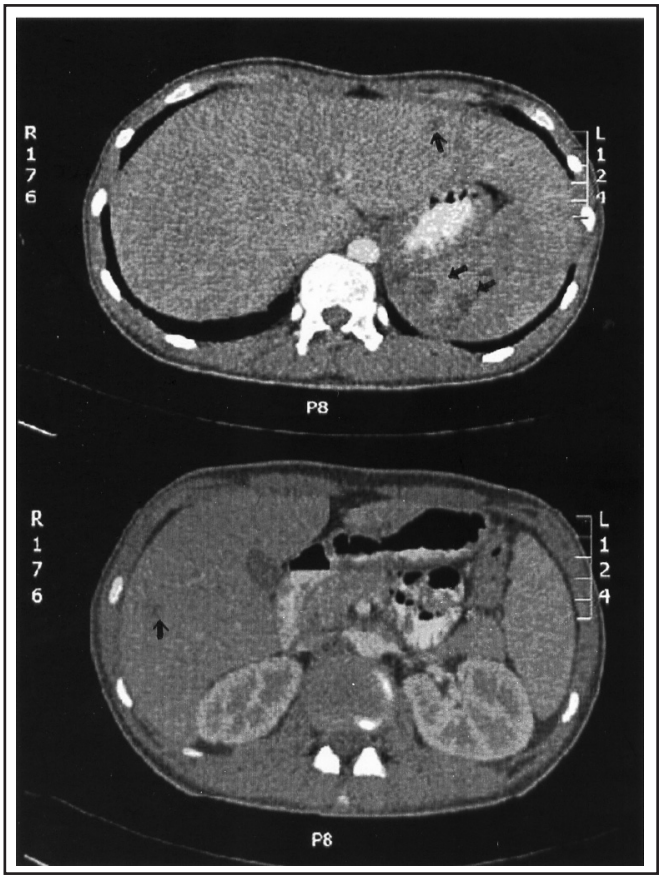

Fig. 1: Multiple ring enhancing lesions seen in the liver and spleen.

reported have ended in deaths[3]. This infection most often occurs after exposure to contaminated water or soil, and the most frequent entry point is skin lesion, as demonstrated in our patient. *Chromobacterium violaceum* infection frequently presents with sepsis often associated with fever, pneumonia, liver, lung, or splenic abscesses. Almost half of the infected patients have positive blood cultures. Less common presentations include orbital cellulitis, osteomyelitis, conjunctivitis, meningitis, or brain abscess[5].

Chronic granulomatous disease (CGD) is a genetically determined primary immunodeficiency disease in which phagocytic cells are unable to kill certain bacteria and fungi after ingesting them due to inability to create the reactive oxygen metabolites that are necessary for efficient intracellular microbicidal activity. It has been reported that patients with CGD are particularly susceptible to *C. violaceum* infection[5]. It is therefore not completely surprising that our immunocompromised patient had contracted the infection eight years earlier even though it is an uncommon infection. However, it is interesting to note that this patient subsequently had two more episodes of infections and survived, despite the high mortality rate of this infection. To our knowledge, this is the first reported case of recurrent infection in an immunocompromised patient. There are only two other recurrent infection cases reported, both involving immunocompetent patients. The optimal treatment for *C. violaceum* is not well established; it is unclear what the most appropriate treatment is, and the optimal duration for antimicrobial agent. However, we believe that the most critical part of the treatment is the timing – appropriate treatment, either antimicrobial agent or surgical drainage, need to be commenced promptly. In our case, due to high awareness of the patient's condition, culture of pus from the septic lesion and sensitivity of the organism was determined urgently and appropriate treatment was commenced without delay. Other authors have concluded that the worst prognosis in *C. violaceum* infection occurred in patients who had bacteremic sepsis and had not promptly received an effective antimicrobial drug[5].

CONCLUSION

C. violaceum can cause severe infection leading to fulminant sepsis, multi-organ system failure and death. Due to its rarity, clinicians often do not appreciate its importance. Since it is a disease of the tropics, it is therefore important for us to be aware of this infection and its clinical spectrum. We believe that with high clinical suspicion, early detection and adequate treatment, it is possible to reduce the mortality associated with this infection, even in immunocompromised patients.

REFERENCES

1. Sneath PH, Whelan JP, Bhagwan SR, Edwards D. Fatal infection by *Chromobacerium violaceum*. Lancet 1953; 265: 276-7.
2. Macher AM, Casale TB, Fauci AS. Chronic granulomatous disease of childhood and *Chromobacterium violaceum* infections in the southeastern United States. Ann Intern Med 1982; 97: 51-5.
3. Bolton BD, Johnson LW. Recurrent nonfatal *Chromobacterium violaceum* infection in a nonimmunocompromised patient. Infect Med 2000; 17: 686-9.
4. Petrillo VF, Severo V, Santos MM, Edelweiss EL. Recurrent infection with *Chromobacterium violaceum*: first case report from South America. J Infect 1984; 9(2): 167-9.
5. Sirinavin S, Techasaensiri C, Benjaponpitak S, Pornkul R, Vorachit M. Invasive *Chromobacterium violaceum* infection in children: case report and review. Pediatr Infect Dis J 2005; 24(6): 559-61.