

Burkholderia gladioli associated abscess in a type 1 diabetic patient

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ABSTRACT

تحدث البكتيريا المسببة للأمراض "قلادبول بوركولديريا" أمراضاً معدية ومختلفة للأشخاص ممن لديهم نقص المناعة. نستعرض هنا ولأول مرة تقريراً لحالة جسم غريب مرتبط بالبكتيريا "قلادبول بوركولديريا"، ذات العلاقة بالخراج لمريض مصاب بداء السكري النمط (1) المؤهل مناعياً. وقد تمت دراسة مطبوعات طبية ذات العلاقة لفهم تواجد هذا الطراز الاستثنائي لهذه الجرثومة لدى مريضنا.

Burkholderia gladioli, a bacterial pathogen, causes a variety of infectious ailments in immunocompromised individuals. We present a case report of foreign body associated *Burkholderia gladioli* related abscess in an immunocompetent type 1 diabetic patient. Relevant medical literature is reviewed to understand this unusual mode of presentation of this bacterium in our patient.

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Since its identification in 1992, many case reports have implicated *Burkholderia gladioli* (*B. gladioli*) as a human pathogen primarily causing infection in immunocompromised individuals.¹⁻⁴ Because in vitro laboratory investigations demonstrate that *B. gladioli* strains are susceptible to complement mediated lysis of pooled human serum, coupled with lack of recognizable virulence associated factors, it is generally believed that healthy individuals should be immune to infection

from this bacterium.⁵ Here, we present for the first time a case report in which *B. gladioli* caused foreign body associated abscess in an "immunocompetent" type 1 diabetic patient.

Case Report. A 17-year-old boy with medical history of type 1 diabetes for 6 years presented to us with diabetic ketoacidosis and right-sided waist swelling of 3 years in duration. The swelling was initially small, but progressively became enlarged especially in the last 2 months causing pain and discomfort. He uses the peri-umbilical region for sub-cutaneous insulin administration, and denied using the waist region for that purpose. He also denied having any history of recurrent infections in the past. On examination he was mildly dehydrated, he was afebrile and vital signs were stable without evidence of orthostatic hypotension. A 7 x 3.5 cm swelling was evident on the right side of the waist (Figure 1). It was soft but tender on palpation and fluctuation could easily be appreciated. The base of the swelling was indurated, and the overlying skin was erythematous. The hemogram with differential cell count was within normal limits. A blood glucose value of 43 mmol/l (774 mg/dl), serum bicarbonate of 16 mmol/l (normal range 21-32 mmol/l), along with positive serum ketones and low arterial pH of 7.30 were all compatible with the diagnosis of mild diabetic ketoacidosis. Needle aspiration of the swelling yielded approximately 15 ml of sanguinous fluid, the culture of which grew *B. gladioli*, which was sensitive to Fluoroquinolones, Gentamicin, and Piperacillin/Tazobactam. The organism was identified using Phoenix automated system for identification and sensitivity (BD¹ Diagnostic Systems, France, Europe). Due to re-accumulation of the fluid, a surgical incision and drainage was carried out during his care and the wound cavity was irrigated with normal saline and packed with sterile gauze. During the procedure, a foreign body was flushed out of the wound, which turned out to be an air gun pellet on closer analysis (Figure 2). At this stage he recalled that 3 years ago while playing with his friends,

shooting at each other with the air gun, he sustained an injury over this site, which healed but left behind a residual bump. The surgical incision subsequently healed with usual wound care and antibiotic treatment with oral levofloxacin.

Discussion. *Burkholderia gladioli*, a non-fermentative gram-negative rod, is primarily a plant pathogen which causes decay of gladiolus plants, onion bulbs, and mushrooms.⁶ The genus *Burkholderia* was previously classified as *Pseudomonads* but because of phenotypic differences between them, many *Pseudomonas* like species are currently transferred to the this new genus including *Burkholderia cepacia*, *Burkholderia pseudomallei*, *Burkholderia mallei*, *Burkholderia caryophylli*, and *Burkholderia gladioli*, and so forth, and the list is ever since a growing one.⁷



Figure 1 - Swelling on the right side of the waist measuring 7 x 3.5 cm.



Figure 2 - Air gun pellet retrieved from patient's swelling.

In humans, *B. gladioli* has a low pathogenicity for immunocompetent individuals, but has been extensively implicated in causing infections such as osteomyelitis, septicemia, pneumonia, and empyema in immunocompromised individuals affected with chronic granulomatous diseases, cystic fibrosis, AIDS, and liver transplant.⁸⁻¹¹ In addition, a case of *B. gladioli* septicemia is reported from iatrogenic inoculation in a type 2 diabetic patient who underwent diagnostic and therapeutic cardiovascular procedures for acute myocardial infarction.¹² It should be noted, however, that due to the high degree of phenotypic similarity between this species and closely related species in the *Burkholderia cepacia* complex, accurate identification is often very difficult. Routine laboratory tests to identify these bacteria include selective culture media, phenotypic methods, and semi-automated or automated commercial systems. In addition methods that have been validated and considered gold standard include polymerase chain reaction (PCR) based assays, whole-cell protein electrophoresis, PCR-restriction fragment length polymorphism, amplified fragment length polymorphism, and ribotyping.¹³ The drawback of most of these specialized methods is that they require advanced skills and equipment that are not routinely available in most of the clinical microbiology laboratories. In our patient, *B. gladioli* was identified using Phoenix automated system, which is reported to have excellent accuracy to correctly identify this organism but does not have similar accuracy for identifying *Burkholderia cepacia* complex.¹⁴

We believe our case report endorses the traditional wisdom that a retained foreign body can compromise the local immune response leading to infection and abscess formation. This is the most likely explanation in our, otherwise, immunocompetent type 1 diabetic patient who presented with foreign body associated *B. gladioli* related abscess. We know foreign bodies, regardless of the site of implant, remain devoid of a microcirculation, which is crucial for host defense and the delivery of antibiotics. In an excellent review on this subject, the authors noted that the presence of subcutaneous foreign body reduces the minimum inoculum of a bacterium that is required to cause infection by a factor of more than 100,000.¹⁵ It has also been observed in experimental studies, that this increased susceptibility to infection is at least partially due to locally acquired granulocyte defect. It is believed that in a paracrine fashion the activated neutrophils on foreign surfaces result in the release of neutrophil derived peptides that deactivate granulocytes.^{16,17} More recently, it has been shown that certain bacteria such as *Staphylococcus epidermidis* and *Staphylococcus aureus* cause foreign body associated infection because of their ability to colonize on the surface of the foreign body by

forming an adherent multilayer cell organization called biofilm.¹⁸ The biofilm represents a protected growth environment for bacteria that renders bacterial cells less susceptible to the normally hostile environment of tissues and blood.¹⁹ We hypothesize that in our patient it was an interplay of all these processes that might have adversely affected the local immune response in the vicinity of the contaminated foreign body, thus favoring abscess formation from *B. gladioli*, a relatively non pathogenic organism. Clearly, the most important way to avoid infection in such patients is to completely remove the foreign body if possible. Any wound that penetrates the skin should be evaluated meticulously to determine if exploration for foreign bodies is needed. Exploring the history of the mechanism of injury and accurate description of the probable object can help localize the problem and determine the need for removal.^{20,21} It is quite possible that the patient may not recall a remote event and the initial injury may go unnoticed, as was the case with our patient, and the retained foreign body becomes symptomatic years later. Therefore, we suggest that any patient presenting with an abscess at an unusual anatomic location should be radiographed for the presence of foreign bodies. The most widely used radiologic modalities include plain radiography, computed tomography (CT), and ultrasonography. We prefer plain radiography as the initial modality of choice for viewing radiopaque foreign bodies, including metals, bones, teeth, pencil lead, certain plastics, glass particles, gravel and stones, and so forth.²² Although CT is more sensitive than radiography, its increased cost limits its use to those foreign bodies that are not visible on initial radiography and pose a risk of infection or joint destruction. Ultrasonography is widely available and helpful in finding wooden or radiolucent foreign bodies.²³

In conclusion, we report a case of *B. gladioli* related abscess caused by a retained foreign body in an immunocompetent type 1 diabetic patient. We learnt an interesting lesson from our patient that an unusual infection at an unusual anatomic site demands extra vigilance and meticulous exploration to avoid pitfalls in diagnosis and treatment.

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