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Tropical pathology

Burkholderia gladioli sinonasal infection

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ABSTRACT

Introduction: *Burkholderia gladioli* are non-fermenting, Gram-negative, rod-shaped aerobic bacteria that were first identified as a plant pathogen. Most of the *B. gladioli* infections reported in the literature have involved immunocompromised adults and newborn infants. *B. gladioli* in humans is often associated with a poor prognosis.
Case report: We describe the first case of sinonasal infection due to *B. gladioli* and Staphylococcus aureus in an immunocompetent patient who had recently travelled to the Congo.
Discussion: As in the few other reported cases involving immunocompetent patients, the appropriate approach to this multidrug-resistant *B. gladioli* infection was a combination of surgery and antibiotics chosen in the light of an antibiogram.

1. Introduction

Burkholderia gladioli are aerobic, non-fermenting, Gram-negative, rod-shaped bacteria that were initially identified as a plant pathogen. Their isolation in humans was first described in the mid-1990s in two patients with cystic fibrosis and chronic granulomatous disease. Most of the *B. gladioli* infections reported since have involved immunocompromised adults and newborn infants. In humans, *B. gladioli* is an opportunistic pathogen often associated with a poor prognosis [1].

B. gladioli have the following microbiological characteristics:

- they produce various β-lactamases;
- they are resistant to many drugs;
- they acquire plasmids harboring antibiotic-resistant genes;
- they form a biofilm on colonized surfaces.

All of these features raise clinical issues for the treatment of the infection, and are associated with unfavorable outcomes [1]. Reporting on 4 cases of *B. gladioli* infection (with bacteremia, pneumonia, and cervical adenitis) in two adults and two young children (three of them severely immunocompromised), Graves (1997) found the strains susceptible to quinolones, aminoglycosides, and imipenem, and also that *B. gladioli* were isolated together with other pathogens [2].

Here we describe the first case of maxillary sinus infection due to *Staphylococcus aureus* and *B. gladioli* in an immunocompetent patient. There has been a growing microbiological/pharmacological interest in this latter pathogen since an antimicrobial activity screen of *B. gladioli* BCC0238 (a clinical isolate from a cystic fibrosis patient) recently led to the discovery of gladiolin, a novel macrolide antibiotic with a powerful activity against *Mycobacterium tuberculosis* [3].

2. Case report

In November 2015, a 47-year-old Caucasian female patient was referred to our otolaryngology unit with a history of several years of nasal obstruction and frontal headache. The patient reported mugwort and wormwood allergy, but no asthma or intolerance of nonsteroidal anti-inflammatory drugs. She also described a craniofacial trauma two years earlier and a recent trip to the Congo. Nasal examination with a 30°/4 mm endoscope showed slight deviation of the nasal septum, and severely hypertrophic and congested nasal mucosa, with bleeding on exploration. There were no polyps, but there were adenoids in the nasopharynx. Treatment was instituted with nasal irrigations of saline solution, mometasone furoate nasal spray (100 μg in each nostril once a day) and oral rupatadine (10 mg, once a day). Two months later, a CT scan of the paranasal sinuses showed left maxillary sinus filling with mucous-purulent exudate containing air bubbles and confirmed the deviation of the nasal septum (Fig. 1). The treatment was changed to: nasal irrigations with saline solution, azelastine hydrochloride and fluticasone propionate nasal spray (137/50 μg in each nostril twice a day), and oral ebastine (20 mg once a day for six months). In September

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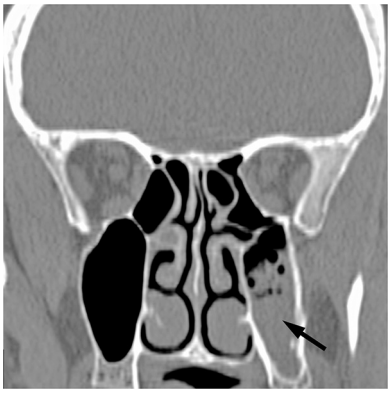

Fig. 1. Coronal view on CT of the paranasal sinuses showing left maxillary sinus filling with mucous-purulent exudate containing air bubbles (arrow).

2016, the patient underwent endoscopic sinus surgery (left ethmoidectomy and middle antrostomy with septoplasty) due to the persistence of symptoms. On discharge, she was prescribed oral amoxicillin-clavulanate (1 g three times a day for 8 days).

Microbiological culture of the intraoperatively sampled purulent secretions from the left maxillary sinus revealed: *B. gladioli* according to the EUCAST (http://www.eucast.org/), which proved resistant to amoxicillin-clavulanate, ertapenem, cefotaxime and ampicillin-sulbactam, but sensitive to levofloxacin (MIC $\leq$ 1 μg/mL); and *S. aureus*, which was sensitive to levofloxacin (MIC < 0.25 μg/mL). Oral levofloxacin was consequently prescribed (750 mg daily for 7 days). Histology of the maxillary sinus mucosa showed chronic inflammation.

Endoscopic follow-up a month after surgery was normal, and the patient complained of no nasal symptoms. Microbiological culture of her nasal secretions showed no pathogen growth. There was no recurrent sinusitis on endoscopic follow-up in May 2017.

3. Discussion

While there is a fair degree of agreement concerning the microbiology of acute sinusitis, the same cannot be said of chronic rhinosinusitis [4]. The bacteria most commonly isolated in chronic rhinosinusitis without nasal polyps are reportedly *S. aureus*, coagulase-negative *Staphylococcus*, and Gram-negative rods (especially *Pseudomonas aeruginosa*) [5]. The *Burkholderia* genus (*Burkholderia cepacia* complex) was also recently isolated in immunocompetent patients with chronic rhinosinusitis, with [6,7] and without polyps [8,9].

Like *B. cepacia* and other opportunists (such as *Pseudomonas* species), *B. gladioli* has repeatedly been isolated in patients with cystic fibrosis and chronic granulomatous disease, and in immunocompromised patients, often in association with other germs [1]. The isolation of *B. gladioli* has been associated with a poor prognosis, and with the need for specific measures to reduce the risk of complications, which may sometimes even be fatal. Judging from the available literature, a period of isolation may be necessary for newborn infants affected, and appropriate surgery with multi-antibiotic therapy for patients with complicated cystic fibrosis. Among patients with cystic fibrosis who have undergone lung transplantation, a higher mortality rate has been described in cases in which *B. gladioli* was isolated [1]. *B. gladioli* infections associated with other germs have rarely been reported in immunocompetent patients. Targeted and timely multi-antibiotic therapy seems to be the key to clinical success [2].

In January 2015, 75 people died and 177 were hospitalized after attending a funeral in the village of Chitima in Mozambique [10]. The deaths were linked to the consumption of a traditional African beverage called *pombe*. Quantitative analysis found potentially fatal levels of the toxic bongkrekic acid in samples of the beverage concerned. Bongkrekic acid is known to be produced by *B. gladioli*. The bacterium could not be isolated from the pombe, but was isolated from corn flour (an ingredient used in the production of pombe) obtained from the brewer's home [10]. Our patient denied drinking any of the local craft drinks while staying in the Congo, however.

This is the first reported instance of *B. gladioli* isolated in an immunocompetent patient with chronic rhinosinusitis without nasal polyps. The clinical behavior of this multidrug-resistant *B. gladioli* infection made it necessary to treat the patient with a combination of surgery and antibiotics (based on the results of an antibiogram).

Disclosure of interest

The authors declare that they have no competing interest.

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