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Case report

Ochroconis gallopava bronchitis mimicking haemoptysis in a patient with bronchiectasis

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ABSTRACT

Ochroconis gallopava is an anamorphic mould characterized by slow growth rate and production of a maroon pigment, which has been isolated worldwide from soil, thermal springs, decaying vegetation, and chicken litter. It has been reported to cause localized, mostly pulmonary, and systemic infection in severely immunocompromised patients.

We describe the case of a 76-year-old woman known for ulcerative colitis-related bronchiectasis treated with low dose oral steroids, who developed a fungal bronchitis with dark, bloody-like, sputum which was initially misinterpreted as haemoptysis. A filamentary mould grew on sputum culture, and was identified by DNA analysis as *Ochroconis gallopava*. We observed a significant clinical improvement after 6 weeks of itraconazole therapy.

1. Case report

A 76-year-old woman, who never smoked, known for ulcerative colitis-related bronchiectasis, reported an increasing quantity of dark sputum and shortness of breath since 3 months. She reported every morning the expectoration of a small cup of dark brownish-red sputum. She denied fever, weight loss or other constitutional symptoms. Further, she reported several self-limited episodes of was describes as non-hypoxemic haemoptysis episodes over the last year. On chest CT scan, the bronchiectasis was progressive over the last 3 years and predominant in the lower lobes. Ulcerative colitis was currently in remission under an oral treatment of mesalazine and prednisolone <10mg/day. The past medical history revealed corticosteroid-induced osteoporosis, peripheral artery occlusive disease, and a previous alcohol abuse. Physical examination showed a slightly malnourished woman in preserved general condition, and normal vital parameters. Lung auscultation revealed humid rales over both lung bases. Clubbing was absent.

The current treatment included prednisolone 7.5mg/day, inhaled budenoside, mesalazine, calcium and vitamin D, azithromycin 250 mg 3 times a week, clopidogrel and levetiracetam.

Lab results showed increased inflammatory parameters (CRP 38 mg/L, leucocytes 12.5 G/L with a slight lymphopenia of 1.3 G/L). Pulmonary functions tests showed normal static and dynamic lung volumes and a mild reduction of carbon monoxide diffusion capacity. Chest X-ray and chest CT-scan showed large cylindrical bronchiectasis with right lower lobe predominance, diffuse bronchial wall thickening, and mucus plugging (Fig. 1). Sputum cytology showed many leucocytes but no erythrocytes or haemosiderophages.

The initial differential diagnosis included a lower respiratory tract bacterial infection, haemoptysis in the context of bronchiectasis and tortuous bronchial arteries, or allergic bronchopulmonary aspergillosis. The latter diagnosis was ruled out by the absence of asthma, normal total IgE levels (47 kU/L), and negative sputum cultures for *Aspergillus*. Other diagnoses such as pulmonary embolism or vasculitis were considered unlikely considering the clinical presentation.

An empirical treatment with moxifloxacine was administered for 14 days without any improvement. Sputum culture showed no growth of bacteria or yeast. However, mould culture grew 34 colonies of a filamentary mould producing a red pigment, which was identified as *Ochroconis gallopava* (also called *Ochroconis gallopavum*) by DNA analysis (Fig. 2). Fungigram showed a minimum inhibitory concentration (MIC) of 0.12 ml/L, 0.06 ml/L and 0.03 ml/L for itraconazole, voriconazole and posaconazole, respectively.

The slow clinical deterioration with increasing bronchorrhea,

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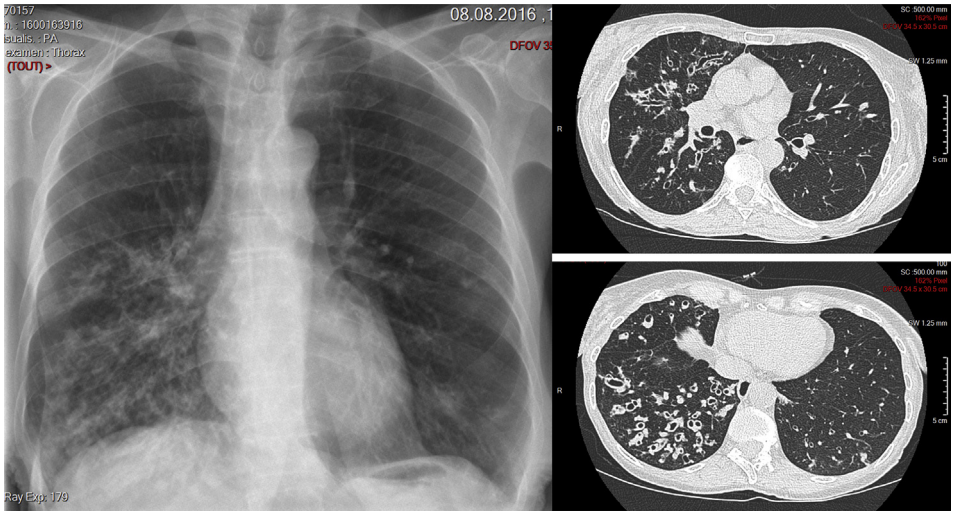

Fig. 1. Chest X-ray and CT scan showing bronchiectasis with air-fluid levels, and bronchial wall thickening with right lower lobe predominance.

the detection of *O. gallopava* in sputum cultures, the absence of other relevant pathogens, and the lack of clinical improvement with antibiotic treatment led to the diagnosis of a fungal bronchitis. Itraconazole 200mg/day was administered orally for 6 weeks with a clinical improvement and disappearance of brownish-red sputum. One month after treatment completion, the sputum culture for mould was sterile.

2. Discussion

This case illustrates the occurrence of acute bronchitis due to *O. gallopava* in an immunocompetent patient with bronchiectasis. So far, only one case of a pulmonary infection caused by this fungus in an elderly immunocompetent host with bronchiectasis was reported [1].

O. gallopava is a melanin producing [2] anamorphic mould characterized by slow growth rate and production of a red to maroon pigment which was initially observed to cause epidemic avian encephalitis, and which was recently transferred to the new genus *Verruconis* [3]. It has been isolated worldwide from soil, thermal springs, decaying vegetation, and chicken litter. It is the most pathogenic species from the genus *Ochroconis* in humans and may causes localized, mostly pulmonary, and systemic infection in immunocompromised hosts [4]. Solid organ transplant patients are particularly at risk for infections by this pathogen and survival is dramatically reduced if brain infection occurs [5]. Although bronchiectasis may predispose to colonisation and infection by fungi, *O. gallopava* as the causal agent has been reported so far in only one case [1]. There is no report of such infection in cystic fibrosis. However, as this organism is poorly known and difficult to identify by conventional cultures, colonisation and infection may be underrecognized. DNA sequencing of the internal transcribed spacer no. 2 of the nuclear rRNA gene operon or of the D2 region of large-subunit ribosomal RNA gene are the gold standard for identification and should therefore be early considered [6].

Our patient presented with brownish-red sputum which was misinterpreted as haemoptysis for several months in the context of bronchiectasis. *Ochroconis* produces a red to marron pigment which may be mistaken for coagulated blood. This case illustrates the interest of sputum analysis to confirm presence of erythrocytes or haemosiderophages before performing invasive diagnostic and therapeutic intervention such as bronchoscopy, or bronchial artery embolization.

We observed a significant clinical improvement after a 6-week treatment with itraconazole in an host treated with low dose steroid and without clinical signs of central nervous system involvement.

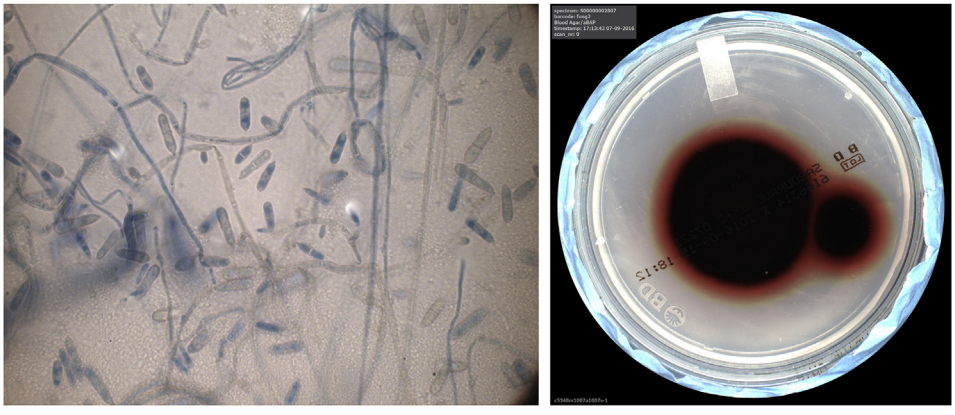

Fig. 2. A and B: Culture of *Ochroconis gallopava*. A, macroscopic aspect of colony on Sabouraud medium after four days of growth at 30 °C showing red to maroon pigment around the colony. B, microscopic aspect of the same colony showing two-celled conidia (lactophenol cotton blue staining). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

There are currently no guidelines regarding the optimal antifungal regimens for *Ochroconis* species. However, several reports suggest that posaconazole and itraconazole may be effective, followed by amphothericin B and voriconazole [5,7–10]. These results are in agreement with *in vitro* susceptibility data [11] as well as the MIC values observed in our case. Early diagnosis and treatment of *O. gallopava* infection is mandatory to avoid dissemination to the brain, which carries a very poor prognosis [12]. The good penetration of posaconazole into the central nervous system and its low MIC well below the serum levels achievable with standard dosing regimens [13] supports its use for difficult-to-treat disseminated brain infections.

In summary, we report the occurrence of an *O. gallopava* acute bronchitis in a patient with bronchiectasis with three relevant features. Firstly, *O. gallopava* may cause symptomatic infection in a patient with bronchiectasis taking only low dose of steroids. Secondly, infection with this mould secreting extracellular red to maroon pigment may mimic haemoptysis. Thirdly, similarly to immunocompromised patients, an azole-based antimycotic therapy may be effective in this clinical context.

Established facts

Ochroconis gallopava may cause pulmonary and systemic infection in severely immunocompromised patients.

Novel insights

Ochroconis gallopava bronchitis may cause dark, bloody-like, sputum which may be misinterpreted as haemoptysis and occur in non severe immunocompromised patients.

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