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Case Report

Pleuropulmonary nocardiosis, an unusual radiological presentation: Case report *

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ABSTRACT

Nocardiosis is caused by strict aerobic filamentous bacteria of the genus Nocardia belonging to the order Actinomycetales with Actinomyces, Streptomyces and Mycobacterium. The radio-clinical presentation in the chest is often misleading. We report a case of pulmonary nocardiosis with an unusual radiological presentation. A 54-year-old patient, chronic smoker, never treated for pulmonary tuberculosis, who presented with a chronic cough complicated by moderate hemoptysis, all evolving in a context of altered general condition and feverish sensations. The radiological aspect was in favor of a hydropneumothorax, the pleural puncture brought back a chocolate colored purulent liquid with the presence of numerous yellow grains and the direct examination showed numerous branched gram-positive bacilli. The bacteriological study allowed to retain the presumptive diagnosis of nocardiosis, the patient was put under antibiotic treatment with a clear clinical and radiological improvement. This observation illustrates the diagnostic difficulty of pulmonary nocardiosis and emphasizes the importance of thinking about nocardiosis in front of any dark thoracic syndrome.

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Introduction

Nocardiosis is a granulomatous and suppurative infection caused by a bacterium of the genus Nocardia, telluric; ubiquitous that belongs to the Actinomycetes group [1]. The species

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Fig. 1 – Chest X-ray on admission showing a dense homogeneous opacity obliterating the left pleural recesses and the left edge of the heart, which took up almost the entire lung, with a hydroaerobic level.

N. asteroides seems to be the most common [2,3]. Pulmonary involvement represents 60%-80% of the described forms [4]. It occurs most often in debilitated patients. The positive diagnosis of pulmonary nocardiosis is difficult. It is pathology due to a pathogen of delicate bacteriological identification with a nonspecific radio-clinical presentation posing the problem of differential diagnosis with bronchopulmonary cancer and certain granulomatoses.

We report a case of pulmonary nocardiosis with an unusual radiological presentation.

Observation

A 54-year-old farmer by trade who has smoked for 46 packyears and has never received treatment for pulmonary tuberculosis presented 10 months prior to admission with a productive cough producing yellowish sputum, occasionally complicated by moderate amounts of hemoptosis, all of which were developing in the context of an altered general condition and feverish feelings. The clinical examination found a patient apyretic at 37.8°C polypneic at 24 cycl /mn, SaO₂ = 89% in the open air and 96% under O₂, a dullness which takes all the left pulmonary hemithorax, bad oral condition and bilateral axillary adenopathies of 1 cm firm mobile without inflam-

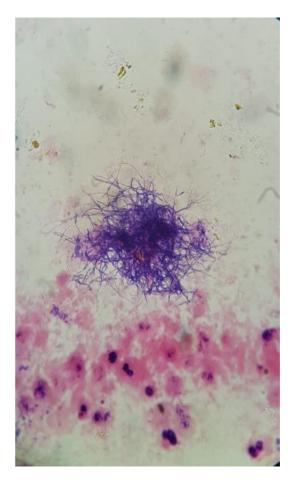


Fig. 2 – Presence of branched filamentous Gram-positive bacilli on direct examination of pleural fluid (objective * 100).

matory signs in front. A chest X-ray was performed showing a hydro-pneumothorax appearance (Fig. 1).

The pleural puncture brought back a chocolate-colored purulent fluid and the direct examination showed numerous filamentous gram-positive bacilli evoking in the first place actinomyces (Fig. 2).

The search for BK and GeneXpert MTB /RIF in the pleural fluid and in the sputum was negative. The biological workup revealed a biological inflammatory syndrome (CRP = 180 mg/L), microcytic hypochromic anemia at 8.1 g/dL, hyperleukocytosis at 16,000/mm³ with a predominance of neutrophils, fasting hyperglycemia at 2.1 g/L. HIV serology was negative.

Chest CT scan showed a left lung abscess communicating with the lobar bronchi of the left stem bronchus, left pyopneumothorax appearing to communicate with the lung abscess and right peribronchovascular micronodules with infectious appearance (Fig. 3).

The laboratory, after the suspicion of actinomycosis, put in culture the pleural liquid in prolonged incubation, after 15 days of incubation in strict aerobic atmosphere (Fig. 4), the culture on agar with cooked blood and on Columbia agar with 5% blood, were positive and allowed the isolation of bacterial strains in favor of Nocardia (Fig. 5): the appearance of the

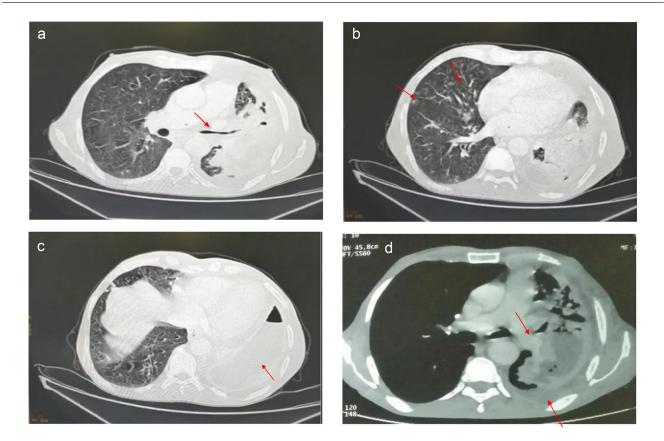


Fig. 3 – (A) Thoracic CT scan with axial parenchymal section in favor of a left pulmonary abscess communicating with the left lobar bronchus. (B) Thoracic CT scan with axial parenchymal section showing peribronchovascular and subpleural pulmonary nodules and micronodules in the right lobar. (C) Thoracic CT scan with axial parenchymal section showing a left pyo-pneumothorax (D): Chest CT scan with mediastinal sections: in favor of a left pulmonary abscess communicating with the left lobar bronchus, with a left pyo-pneumothorax appearing to communicate with the pulmonary abscess.

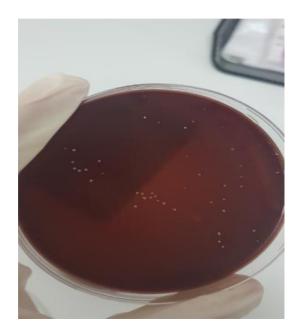


Fig. 4 – Pure positive culture on cooked blood agar after 15 days of aerobic incubation.

colonies (color of the young colonies in white, presence of hyphae in periphery, slightly raised, tendency to become embedded in the agar with time) The realization of Gram staining (variable aspect: bacilli, filaments, coccobacilli) and of modified kinyoun (presence of a partial acido-alcohol resistance), allowed to make a presumptive identification of genus Nocardia and to rule out the genus Actinomyces. Identification at species level was not made by molecular biology PCR 16 S due to the nonavailability of this technique and the antibiotic sensitivity test was not performed due to technical difficulties in isolating the strain.

The patient was put on Amoxicillin + clavulanic acid injection at a rate of 3 g per day, the evolution was marked after 2 weeks of treatment by a clear clinical improvement (reduction of sputum, apyrexia and stop of hemoptysis) biological (CRP control at 56 mg/L and white blood cells at 12,000) and radiological (Fig. 6).

Discussion

Nocardiosis are caused by strict aerobic filamentous bacteria of the genus Nocardia belonging to the order Actinomycetales with Actinomyces, Streptomyces and Mycobacterium [4]. Are

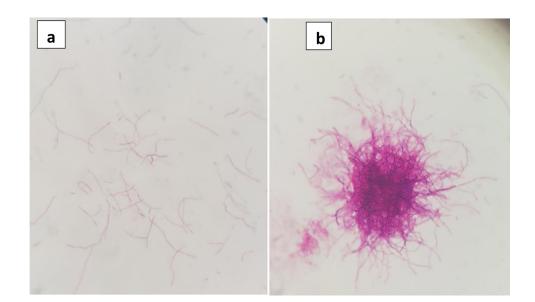


Fig. 5 – (A)Gram stain from culture (cooked blood agar) showing branched filamentous bacilli, (B) image a with magnification.



Fig. 6 – control chest X-ray after 2 weeks showing the beginning of radiological cleaning.

hydrotelluric bacteria widely distributed in the environment, living in a saprophytic state in the soil and many species are pathogenic to humans and animals and plants.

The frequency of pleuropulmonary involvement varies from 44.3% to 85%, explained by the most frequent mode of contamination: inhalation of fragments of filaments or spores present in the air, especially in dust [5]. Nocardiosis remains a pathology of immunologically fragile subjects, especially HIV positive subjects, long term corticosteroid therapy, transplant patients, carriers of solid neoplasia or hematological malignancy, hepatic and renal diseases [6] as well as cardiac pathologies and autoimmune hemolytic anemia [7]. In our patient, smoking and inaugural diabetes were factors that favored this infection

The most frequent clinical form is a pulmonary infection (present in 60%-80% of cases) [8,9]. Its diagnosis is difficult, especially in cases of isolated pulmonary involvement. The clinical picture is generally aspecific, with a slow respiratory symptomatology that is resistant to the usual antibiotic treatments, leading to suspicion of tuberculosis, mycoses or neoplasia [10,11]. This is the case of our patient in whom the diagnosis of pulmonary nocardiosis was only confirmed after 8 months of evolution. The clinical manifestations are variable: anorexia, weight loss, productive cough, dyspnea and sometimes hemoptysis [11]. Various radiological abnormalities can be found: nodules or localized or disseminated foci, interstitial or reticulonodular infiltrates or even miliary aspects, pulmonary condensations, pleural effusions, mediastinal adenopathies [12-14]. In our patient, the radiological presentation was unusual with a hydropneumothorax aspect.

The differential diagnosis of pulmonary Nocardiosis is multiple and is essentially made with Actinomycosis, tuberculosis, pulmonary cancer, Wegener's granulomatosis and pulmonary sequestration. Direct examination by light microscopy after staining, as a simple rapid examination, can easily differentiate between infectious and non-infectious pathologies especially in favor of Nocardia /Actinomyces /Mycobacteria.

The positive diagnosis of Nocardiosis is difficult. It is a pathogen of delicate bacteriological identification that requires a confirmatory identification at the genus level by molecular biology using 16 S ribosomal DNA gene sequencing (reference technique). In our patient, the diagnosis of pulmonary nocardiosis was retained on the bacteriological study of the pleural fluid after a direct examination and a positive culture.

The choice of probabilistic treatment will be conditioned by the dissemination of the infection (whether or not the CNS is involved), the existence of severe clinical signs and the patient's immune status. Moreover, as the antibiotic sensitivity profile is unpredictable, the choice of broad-spectrum molecules against Nocardia is essential, pending identification and antibiogram.

In this context, sulfonamides remain the reference treatment [15]. They have good tissue penetration, including brain tissue. Amikacin, meropenem, imipenem, third generation cephalosporins (cefotaxime or ceftriaxone), ciprofloxacin, linezolid or minocycline by parenteral route represent therapeutic alternatives (in case of intolerance or contraindication to sulfonamides [16–18]. Some authors recommend the combination of cotrimoxazole and third-generation cephalosporin, cotrimoxazole-meropenem [19], imipenem-amikacin [16,11], ampicillin-erythromycin or ampicillin-cotrimoxazole [10]. The parenteral route should be recommended. If the course is favorable, oral therapy (usually 4-6 weeks) with cotrimoxazole, minocycline or a fluoroquinolone is recommended [10].

Treatment must be prolonged because of the high risk of relapse [20]. It is 6 months for isolated pulmonary involvement and up to 1 year in case of systemic nocardiosis, cerebral localization or nocardiosis in immunocompromised patients [10].

Our patient was put on Amoxicillin + clavulanic acid, which is an active molecule on certain nocardia, notably asteroides, the most frequently isolated in human medial pathology, with a good evolution.

The main complication of this pulmonary contamination is the secondary dissemination by hematogenous route, responsible for systemic nocardiosis. The most frequent site affected in this case is the nervous system, with the formation of one or more brain abscesses, more rarely meningitis. In the case of multiple localizations, mortality exceeds 50%. Other sites of predilection are the skin, subcutaneous tissue, soft tissue, bones and joints [21]. Its prognosis is conditioned by early diagnosis and treatment.

Conclusion

Pulmonary nocardiosis is a rare disease most often unrecognized. This leads to frequent diagnostic and therapeutic delays with a more or less mutilating and especially inappropriate treatment. This observation illustrates the diagnostic difficulty of pulmonary nocardiosis and emphasizes the interest of thinking about nocardiosis in front of any dark thoracic syndrome.

Patient consent

The authors of this manuscript have obtained written, informed consent from the patient to write up the case report and for the use of images pertinent to the case. We have ensured anonymity of all clinical and graphical data used.

REFERENCES

Granier F. Abcès cérébral à Nocardia farcinica associé à une embolie pulmonaire chez une patiente immunocompétente. Presse Med 2005;34:522–4.

- [2] Bonnet F, Donay J-L, Fieux F, Marie O, de Kerviler E, Jacob L. Nocardiose à Nocardia otitidiscaviarum: pièges et retard de diagnostic. Ann fr d'anesthésie et de réanimation 2007;26:680–4.
- [3] Soraa N, Arsalane L, Ouhdouch Y, Louzi L. Abcès cérébral à Nocardia : A propos d'un cas. Rev Tun Infectiol 2009;2:29–33.
- [4] Beaman BL, Beaman L. Nocardia species: host-parasite relationships. Clin Microbiol Rev 1994;7(2):213–64.
- [5] Le Coustumier EM, Denes E, Martin C, Weinbreck P. Nocardiose: analyse rétrospective d'une série de 19 cas. Rev Med Interne 2016;38(2):81–9.
- [6] Kurahara Y, Tachibana K, Tsuyuguchi K, Akira M, Suzuki K, Hayashi S. Pulmonary nocardiosis: a clinical analysis of 59 cases. Respir Investig 2014;52(3):160–6.
- [7] Mootsikapun P, Intarapoka B, Liawnoraset W. Nocardiosis in srinagarind hospital, Thailand: review of 70 cases from 1996-2001. Int J Infect Dis 2005;9(3):154–8.
- [8] Arduino RC, Johnson PC, Miranda AG. Nocardiosis in renal transplant recipients undergoing immunosuppression with cyclosporin. Clin Infect Dis 1993;16:505–12.
- [9] Kontoyiannis DP, Ruoff K, Hooper DC. Nocardia Bacteremia. Report of 4 cases and review of the literature. Medicine 1998;77:255–67.
- [10] Lerner PI. Nocardiosis. Clin Infect Dis 1996;22:891–905.
- [11] Lerner PI. Nocardia species. In: Mandell GL, Bennett JE, Dolin R, editors. Mandell, Douglas and Bennett's principles and practice of infectious diseases. New York: Churchill Livingstone; 1995. p. 2273–80. Vol 2.
- [12] Sarcinelli-Luz B, Marchiori E, Zanetti G, Mauro Mano C, Abdalla F, Carvalho JF, et al. Pulmonary nocardiosis in the acquired immunodeficiency syndrome computed tomographic findings: a case report. Cases J 2009;2:6642.
- [13] Beaman BL, Beaman L. Nocardia species: host parasite relationships. Clin Microbiol Rev 1994;7:213.
- [14] Hwang JH, Koh WJ, Suh GY. Pulmonary nocardiosis with multiple cavitary nodules in a HIV-negative immunocompromised patient. Intern Med 2004;43:852.
- [15] Smego RA Jr, Moeller MB, Gallis HA. Trimethoprimsulfamethoxazole therapy for Nocardia infections. Arch Intern Med 1983;143:711.
- [16] Mc Neil MM, Brown JM. The medically important aerobic Actinomycetes: epidemiology and microbiology. Clin Microbiol Rev 1994;7:357–417.
- [17] Jerome AL, Paul EB, Todd CL, Wayne LG. Brain and lung lesions in an immunocompromised man. CMAJ 2011;183(5):573–6.
- [18] Kobayashi N, Sueoka-Aragane N, Naganobu N, Umeguchi H, Kusaba K, Nagasawa Z, et al. Disseminated Nocardiosis caused by Nocardia concava with acute respiratory failure and central nervous system involvement treated with Linezolid. Intern Med 2012;51:3281–5.
- [19] Sorrell TC, Mitchell DH, Iredell JR. Nocardia species. In: Mandell GL, Bennett JE, Dolin R, editors. Mandell, Douglas and Bennett's principles and practice of infectious diseases. Philadelphia (PA): Churchill Livingstone Elsevier; 2010. p. 3199–207. Vol 2.
- [20] El Hymer W, Skoumi M, Aniba KH, Ghannane H, Idmoussa A, Tali A, et al. Nocardia brain abscess-Case report and review of littérature. Afr J Neurol Sci 2011;30(2):82–6.
- [21] Couraud S, Houot R, Coudurier M, Ravel AC, Coiffier B, Souquet PJ. Infections pulmonaires à nocardia. Rev Mal Respir 2007;24(3 Pt 1):353–7.