Case report

Q fever presenting as myocarditis

Rania Hammami, Amine Bahloul, Salma Charfeddine, Wiem Feki, N. Ben Ayed, L. Abid, Samir Kammoun

Cardiology Department, Hedi Chaker Hospital, Sfax, Tunisia
Radiology Department Hedi Chaker Hospital, Sfax, Tunisia
Bacteriology Department, Habib Bourguiba Hospital, Sfax, Tunisia
Research Unit UR 17ES37, University of Medicine, Sfax, Tunisia

ABSTRACT

We report the case of a 19-year-old healthy adolescent, living in an urban area, admitted because of acute chest pain and extensive anterior ST elevation. Coronary arteries were normal on coronary angiography; troponins were very high, echocardiography revealed a preserved global systolic function but an alteration of the longitudinal strain in the inferolateral wall. Cardiac MRI confirmed the diagnosis of acute myocarditis. As part of the etiological workup, Coxiella burnetii serology showed an acute infection. The diagnosis of Coxiella burnetii myocarditis was retained and the patient was treated with doxycycline and corticosteroid therapy. The myocardial localization of this germ is unusual but can be serious, hence the interest of a Coxiella serology in endemic countries face to any acute myocarditis.

© 2021 Published by Elsevier Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

Q fever is a zoonosis caused by intracellular germ Coxiella burnetii. Since the discovery of the first case in Africa by Kaplan et al. [1] in 1955, this pathology has been increasingly diagnosed in this continent thanks to the development of bacteriological investigation tools. The prevalence of Q fever in Maghreb countries is largely underestimated [2]. Infection is asymptomatic in 60% of cases [3,4]. Cardiac impairment generally affects endocardium especially in chronic forms, Coxiella serology is not recommended in the systematic etiological assessment of myocarditis.

Coxiella myocarditis is exceptional with an incidence inferior to 0.5% (5.6) and limited to few isolated cases reported in the literature. We described here, a case of acute myocarditis, incidentally revealing a Q fever in a young patient without typical manifestations of infection but living in an epidemic area.

Case report

A 19-year-old, living in an urban area, with neither medical antecedents nor contact with animals was admitted in May 2019 for constrictive chest pain associated with an ST segment elevation in anterior EKG leads. Coronary angiogram revealed normal coronaries. During the interrogation, the patient disproved the notion of taking drugs, fever or influenza syndrome. Transsthoracic echocardiography, showed hypokinesia of the infero-lateral wall with a normal global systolic function. However the overall longitudinal strain was low (-14%, normal: -18 – 22%) especially in hypokinetic segments where regional strain was estimated at -8% (Fig. 1). The peak of troponins I was at 24 g/L (normal: 0.01 g/L), there was a biological inflammatory syndrome (CRP at 150 mg/L, white blood cells at 18,000/L). Cardiac MRI showed a left ventricle ejection fraction at 55% with an inferior epicardial enhancement (Fig. 2). As part of the etiological assessment of acute myocarditis confirmed by MRI and the inflammatory syndrome, different investigations were conducted showing acute Q fever (Table 1). The level of anti-nuclear antibodies was high at 1:1280. Anti-phospholipid antibodies were negative.

The patient was treated with doxycycline 200 mg/day for 45 days. Two-month echocardiography showed a stable ejection fraction, with a worsening of the overall longitudinal strain in the inferolateral territory, the progression to dilated cardiomyopathy was feared. A cardiac MRI showed a persistence of epicardial enhancement. Repeat serology reversed the transition to chronicity (Table 1). In order to avoid myocardial fibrosis, long-term corticosteroid therapy was introduced (high dose for 6 weeks with gradual reduction over 6 weeks). The evolution was favorable, the echocardiography control at 12 months showed a total normalization of the overall longitudinal strain in the different segments (Fig. 1). Cardiac MRI, performed at 12 months, did not show late enhancement.
Discussion

Q fever is a zoonosis, first described in 1937 in Australia [3], is transmitted to humans accidentally by the secretions of domestic animals during parturation or by the consumption of unpasteurized milk. The forms of the bacterium can survive in the air for long periods and can be driven by wind for long distances; direct contact with animals is not required for contamination. In our patient, there was no history of contact with animals neither of unpasteurized dairy products or of living within several miles of a farms where animal parturition occurred.

Clinical presentation is often benign, severe forms depend on the virulence of the strain in question and the host terrain [4]. Infectious endocarditis is the most frequently reported heart damage, and is often seen in chronic infection with high phase 1 antibody and here and elsewhere. In the countries of Maghreb, this germ [4] causes 1–3% of endocarditis. Myocarditis is exceptional, published in isolated cases [5–8], less than 30 cases of myocarditis with Coxiella have been reported; this impairment is certainly underestimated especially in endemic areas.

In our patient, the revelation of the infection was fortuitous conducted be a systematic etiological assessment of myocarditis.

Table 1
Serology of Coxiella burnetii in our patient (seroconversion and no signs of chronicity).

|                | May 08, 2019 | May 25, 2019 | June 7th, 2019 | October 27, 2019 |
|----------------|--------------|--------------|----------------|-----------------|
| IgG phase II   | 1:200        | 1:200        | 1:6400         | 1:1600          |
| IgM phase II   | 1:50         | 1:50 (positive) | 1:50 (positive) | 1:50            |
| IgG phase I    | –            | –            | –              | 1:200           |

![Fig. 1](image1.png) (A) Global longitudinal strain at admission, low especially in inferolateral. (B) Recovery of the global longitudinal strain at 12 months in different regions.

![Fig. 2](image2.png) (A) FIESTA sequence in small axis view and 4 chambers view: hypersignal under epicardial of the lower and lateral wall of the left ventricle. (B) late enhancement sequence: under epicardial and not systematized enhancement particulary in the lateral wall.
In fact the prevalence of Coxiella burnetii infection is high in our country, 26 % of blood donors [2], have a positive serology. Signs of myocarditis are not very specific, making diagnosis difficult and sometimes late in the stage of cardiac complications, including dilated cardiomyopathy and rhythm disorders.

The pathophysiological mechanism of myocardial impairment is poorly elucidated, an intense autoimmune response has been suggested [4], the significant rise in anti-nuclear antibody levels in our patient, without other causes, testified to this autoimmune reaction; direct evidence of the germ in myocardial biopsies has been reported in one patient in the literature, suggesting a direct pathogenicity of this germ [4].

The prognosis of myocarditis is often not clear, due to delayed diagnosis and unspecific clinical presentation. Mortality from Q fever is estimated at 1–2 % [3] while it exceeds 25 % for myocarditis [6].

In the lack of consensus, doxycycline is considered the reference treatment in Coxiella burnetii infections. In case of valve damages, some physicians recommend combining hydroxychloroquine to prevent the transition to chronicity. Some cases of patients treated with levofloxacin or azithromycin with favorable reports have been reported in the literature [7,8]. In our patient after the doxycycline treatment and given the rise of antinuclear antibody levels and the lack of improvement of the global longitudinal strain, we feared myocardial fibrosis and the progression to dilated cardiomyopathy, thus corticosteroid therapy was prescribe beyond the antibiotic therapy period and the evolution was favorable with restitution ad integrum.

Conclusion

Given the endemic feature of Coxiella Burnetti infection in some countries, the improved means of bacteriological diagnosis, the severity of myocarditis with this germ and the unpredictable evolution, the systematic search for this infection would be necessary face to any acute myocarditis case, in these countries.

Sources of funding

We declare no source of funding

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

Ethical approval

Ethical approval for the study was obtained from the ethic committee of Hedi Cheker hospital, Sfax, Tunisia.

Author contribution

RH: wrote the paper
AB, SC, LA and SK: revised the paper
WP: performed and analysed the imaging for the patient
NBA: performed the bacteriologic test.
All the authors have read and agreed to the final manuscript.

Declaration of Competing Interest

The authors do not declare any conflict of interest

References

[1] Kaplan MM, Bertagna P. The geographical distribution of Q fever. Bull World Health Organ 1955;13(5):829–60.
[2] Letaief AO, Yacoub S, Dupont HT, et al. Seroepidemiological survey of rickettsial infections among blood donors in central Tunisia. Trans R Soc Trop Med Hyg. 1995;89(3):266–8.
[3] Raoult D, Marrie T, Mege J. Natural history and pathophysiology of Q fever. Lancet Infect Dis 2005;5(April 4):219–26.
[4] Eldin C, Mélenotte C, Medennikov O, et al. From Q fever to Coxiella burnetii infection: a paradigm change. Clin Microbiol Rev 2017;30(1):115–90.
[5] Meletotte C, Protopopescu C, Million M, et al. Clinical features and complications of Coxiella burnetii infections from the French national reference center for Q fever. JAMA Netw Open 2018;1(4). e181580-e181580.
[6] Fournier PE, Etienne J, Harle JR, et al. Myocarditis, a rare but severe manifestation of Q fever: report of 8 cases and review of the literature. Clin Infect Dis Off Publ Infect Dis Soc Am 2001;32(10):1440–7.
[7] Steffen J, Bogner J, Huber BC. Q-fever - a rare cause for myocarditis. Dtsch Med Wochenschr 1946 2020;145(7):484–7.
[8] Jacobson A, Sutthiwat P. Myocarditis: A rare manifestation of acute Q fever infection. J Cardiol Cases 2019;20(2):45–8.