Joint Bone Spine 86 (2019) 115–116

Available online at
ScienceDirect
www.sciencedirect.com

Elsevier Masson France
EM|consulte
www.em-consulte.com/en

Letter to the Editor

Non-infective endocarditis: Expanding the phenotype of giant cell arteritis

ARTICLE INFO

Keywords:
Giant cell arteritis
Polymyalgia rheumatica
Non-infective endocarditis

A 80-year-old European woman, with no remarkable medical history, was referred in October 2013 for pain in the hips, shoulders, and unusual headaches. She was diagnosed with polymyalgia rheumatica. Prednisone was started at a dose of 0.3 mg/kg/day. The pain disappeared quickly, and three months later, the patient discontinued her therapy, with no more follow-up. In October 2015, she consulted for dyspnoea, and faintness, without symptoms of polymyalgia rheumatica or giant cell arteritis (GCA). A CT-scan revealed an ascending thoracic aorta aneurysm (ATAA) measuring 60 mm in diameter. The patient was referred to a thoracic surgery centre. Trans-thoracic echocardiography confirmed ATAA, associated with aortic insufficiency. The three aortic valve' cusps were thin, with no additional imaging. Aortic insufficiency was due to an annular dilation. Bentall surgery was performed, associated with aortic valve replacement through bioprosthesis. Pathological examination of operative pieces showed an aortitis with giant cells in the media associated with a fragmentation of the internal elastic membrane, which led to the diagnosis of GCA (Fig. 1). The aortic valve examination showed no macroscopic abnormalities, however, endocarditis lesions with intimal infiltration of polymorphonuclear and lymphoplasmacytic cells were observed. Gram and Giemsa stains were negative (Fig. 2). Blood cultures were negative. Searches for HIV, HCV, HBV, *Rickettsia, Treponema, Bartonella*, and *Brucella* were negative. Anti-nuclear antibodies, and anti-neutrophil cytoplasmic antibodies were negative. There were no arguments for an antiphospholipid syndrome. Diagnosis of non-infective endocarditis (NIE), associated with GCA, was then retained. The short-term outcome was notable for a long postoperative stay in intensive care unit due to bilateral diaphragm palsy, transitorily needing mechanical ventilation, potentially related to GCA [1]. Prednisone was restarted at 0.7 mg/kg/day in January 2016. Evolution was good. In May 2017, she did not have any signs of GCA or polymyalgia rheumatica after being treated with 5 mg of prednisone per day. An incomplete bilateral diaphragm palsy was still present with a dyspnoea, however, she did not need respiratory assistance. The patient did not present with any heart complications.

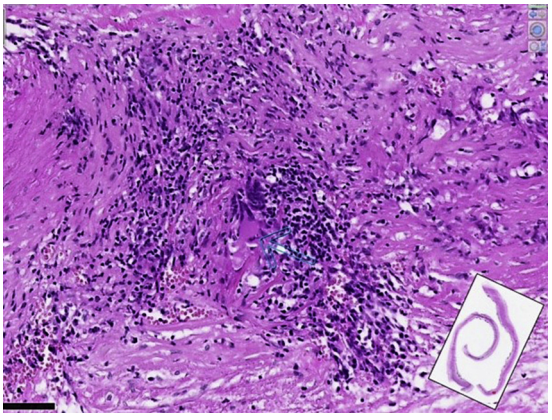

Fig. 1. A microscopic examination of the aorta showing the inflammatory infiltration of the media. The blue arrow shows a giant cell in contact with a fragmented elastic fibre, which is partially destroyed. It also shows a perivascular lymphocytic infiltration around the vasa-vasorum.

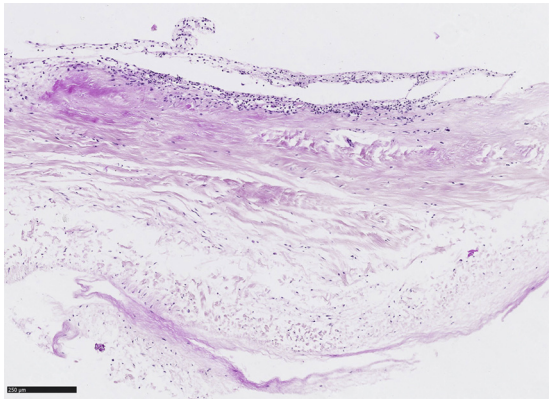

Fig. 2. A microscopic examination of the aortic valve showing a polymorphonuclear infiltration in touch with the endothelial cells of the intima. There is no sign of necrosis, however fibrous remodeling.

To the best of our knowledge, this patient presents with the second case of pathologically proven non-infective endocarditis associated with GCA [2]. Aetiologies of NIE include systemic lupus erythematosus, antiphospholipid syndrome, ANCA vasculitis, spondylarthritis [3–5]. Association between NIE and GCA has been described in three cases, in which endocarditis was defined by vegetation images on echocardiography [6–8], however one of the cases had prostatic cancer, known to be a cause of marantic endocarditis [8]. In our case, as well as in the other pathologically proven one [2], there was no sign of vegetations. Inflammation of the aortic valve could be secondary to the aortitis, however in the other pathologically proven case [2] there was no sign of aortitis. Therefore, NIE could be a specific complication of GCA, even

https://doi.org/10.1016/j.jbspin.2018.04.002

without aortitis. Further observations are needed to confirm this association, and to assess its prognostic value.

Disclosure of interest

The authors declare that they have no competing interest.

References

[1] Blaise S, Liozon E, Nadalon S, et al. Horton's disease revealed by brachial C5 plexopathy. Rev Med Interne 2005;26:578–82.
[2] Niclauss L, Letovanec I, Chassot PG, et al. Acute aortic valve insufficiency and cardiogenic shock due to an isolated giant cell inflammation of the aortic valve leaflets: case report and review of the literature. J Heart Valve Dis 2008;17:343–7.
[3] Lee JL, Naguwa SM, Cheema GS, et al. Revisiting Libman-Sacks endocarditis: a historical review and update. Clin Rev Allergy Immunol 2009;36:126–30.
[4] Anthony DD, Askari AD, Wolpaw T, et al. Wegener granulomatosis simulating bacterial endocarditis. Arch Intern Med 1999;159:1807–10.
[5] Gensler LS. Axial spondyloarthritis: the heart of the matter. Clin Rheumatol 2015;34:995–8.
[6] Eftychiou C, Fanourgiakis P, Vryonis E, et al. Factors associated with non-bacterial thrombotic endocarditis: case report and literature review. J Heart Valve Dis 2005;14:859–62.
[7] Kopterides P, Moyssakis I, Margos P, et al. Echocardiographic findings in patients with temporal arteritis: apropos of one case of temporal arteritis-associated verrucous (Libman-Sachs) endocarditis. Clin Exp Rheumatol 2006;24:S35–7.
[8] Hesselink DA, van der Klooster JM, Schelfhout LJDM, et al. Non-bacterial thrombotic (marantic) endocarditis associated with giant-cell arteritis. Eur J Intern Med 2001;12:454–8.

Alexandre Terré[a,*]
Olivier Lidove[a]
Odette Georges[b]
Patrick Mesnildrey[c]
Hélène Chennebault[a]
Jean-Marc Ziza[a]
[a] *Department of internal medicine, Croix Saint-Simon hospital, 75020 Paris, France*
[b] *Centre of pathology, Bercy, 75012 Paris, France*
[c] *Department of thoracic surgery, Centre cardiologique du nord, 32–36, rue des Moulins Gémeaux, 93207 Saint-Denis cedex, France*

[*] Corresponding author. Department of internal medicine, Croix Saint-Simon Hospital, 125, rue d'Avron, Paris 75020, France.
E-mail address: alexandre.terre.at@gmail.com (A. Terré)

Accepted 18 April 2018
Available online 27 April 2018