Autoimmunity Reviews 15 (2016) 82–92

Contents lists available at ScienceDirect

Autoimmunity Reviews

journal homepage: www.elsevier.com/locate/autrev

Review

Thymoma associated with autoimmune diseases: 85 cases and literature review

C. Bernard [a,*,1], H. Frih [a,1], F. Pasquet [b], S. Kerever [c,d,e], Y. Jamilloux [a], F. Tronc [f], B. Guibert [g], S. Isaac [h], M. Devouassoux [i], L. Chalabreysse [j], C. Broussolle [a], P. Petiot [k], N. Girard [l,1], P. Sève [a,1]

[a] Department of Internal Medicine, Croix-Rousse Hospital, Hospices Civils de Lyon, Claude Bernard University Lyon 1, Lyon, France
[b] Department of Internal Medicine - Oncology, Desgenettes Instruction Hospital of Army, 108 Bd Pinel Lyon 69003, Lyon, France
[c] Department of Anaesthesiology and Critical Care, Lariboisière University Hospital, AP-HP, Paris, France
[d] ECSTRA Team, CRESS, Epidemiology and Statistics Center, Sorbonne Paris Cité, UMR 1153, INSERM, Paris, France
[e] University Denis Diderot - Paris VII, Paris, France
[f] Department of Thoracic and Cardio-vascular surgery, Louis Pradel Hospital, Hospices Civils de Lyon, Bron, France
[g] Department of Thoracic and Cardio-vascular surgery, Lyon Sud Hospital, Hospices Civils de Lyon, Pierre-Bénite, France
[h] Department of Pathology, Lyon Sud Hospital, Hospices Civils de Lyon, Pierre-Bénite, France
[i] Department of Pathology, Croix-Rousse Hospital, Hospices Civils de Lyon, Claude Bernard University Lyon 1, Lyon, France
[j] Department of Pathology, Louis Pradel Hospital, Hospices Civils de Lyon, Bron, France
[k] Department of Neurology, Croix-Rousse Hospital, Hospices Civils de Lyon, Claude Bernard University Lyon 1, Lyon, France
[l] Department of Respiratory Medicine, Louis Pradel Hospital, Hospices Civils de Lyon, Bron, France

ARTICLE INFO

Article history:
Received 16 August 2015
Accepted 20 September 2015
Available online 25 September 2015

Keywords:
Thymoma
Autoimmune disease
Myasthenia gravis
Isaac's syndrome
Morvan syndrome
Thymectomy

ABSTRACT

Objectives: To describe the clinical features, treatment, and outcome of autoimmune diseases (AD) in a cohort of patients with thymoma.
Design: Pathological records from three university hospitals, between 2005 and 2011, were reviewed to identify patients with thymoma. Patients with thymoma and AD were compared with patients with thymoma without AD.
Results: 47/85 (55%) cases of thymoma had AD, including myasthenia gravis (MG) (n = 33), Hashimoto's thyroiditis (n = 4), Isaac's syndrome (n = 3), Morvan syndrome (n = 2), pure red cell aplasia (n = 2), systemic lupus (n = 2), lichen planus (n = 2), and one case of each following conditions: aplastic anemia, autoimmune hemolytic anemia, Good's syndrome, pemphigus, autoimmune hepatitis, Graves' disease, limbic encephalitis, and inflammatory myopathy. Six patients (7%) presented at least 2 ADs. The median duration of follow-up after surgery was 60 months (40–78 months). In 32 patients, the diagnosis of AD preceded the diagnosis of thymoma, in 9 patients, thymoma was diagnosed at the same time as the AD and 7 patients had been operated on when they developed an AD. We found a significative difference on the Masaoka stage between the MG patients and the patients who present another AD (p = 0.028). No risk factor for developing an AD after thymectomy was identified.
Conclusions: We describe here the long-term follow-up of a large series of AD related to thymoma. Our results confirm previous data concerning AD occurrence in patients with thymoma and suggest that preexisting autoimmunity is not a risk factor for developing autoimmune manifestations after thymectomy.
© 2015 Elsevier B.V. All rights reserved.

Contents

Abbreviations: AA, Aplastic anemia; AD, Autoimmune diseases; AIDS, Acquired immune deficiency syndrome; AIHA, Autoimmune hemolytic anemia; AIRE, Autoimmune regulator; APECED, Autoimmune polyendocrinopathy candidiasis ectodermal dystrophy; Casp 2, Contactin-associated protein 2; Lgi 1, Leucin-rich glioma-inactived protein 1; LRP4, Lipoprotein receptor-related protein; MG, Myasthenia gravis; PRCA, Pure red cell aplasia; SLE, Systemic lupus erythematosus; VGKC, Voltage-gated potassium channels.

* Corresponding author at: Department of Internal Medicine, Croix-Rousse Hospital, 103 grande rue de la Croix-Rousse, 69004 Lyon, France. Tel: +33 4 26 73 26 28.
E-mail address: claire.bernard32@gmail.com (C. Bernard).
[1] Contributed equally.

http://dx.doi.org/10.1016/j.autrev.2015.09.005
1568-9972/© 2015 Elsevier B.V. All rights reserved.

1. Introduction

Thymus is a primary lymphoid organ whose main function is to educate T cells. Positive and especially negative selection of T cells takes place in the thymus, where this ensures the acquisition of central T cell tolerance [1,2].

Thymomas are uncommon neoplasias derived from the epithelial cells of the thymus; however, it is the most common mediastinal mass in adults, representing up to 50% of anterior mediastinal masses [3]. The association between autoimmune diseases (AD) and thymoma has long been known [4], especially myasthenia gravis (MG), which has been associated with thymoma in up to 44% of the patients [5]. Conversely, 75–80% of the patients with MG have thymus abnormalities, including thymic hyperplasia (85%) and thymoma in 15–20% [6]. Retrospective studies describing thymectomy outcome in MG have found positive association between surgery and MG improvement with MG remission after surgery [7–9]. Thymectomy is advocated for all MG patients with thymoma, and for some subsets of patients with moderate to severe generalized and seropositive MG without thymoma [10,11]. Besides MG, thymoma is also associated with other AD, mostly described in reports, including pure red cell aplasia (PRCA), systemic lupus erythematosus (SLE), polymyositis, and Good's syndrome [5,12,13]. The impact of thymectomy on the outcome of AD other than MG is variable, and in some cases, thymectomy may worsen the evolution of the disease [14,15]. Little is known about the physiopathology of the association of thymoma and AD, why some patients develop AD after thymectomy, if there are some patients at risk and how to monitor them.

We reviewed AD in a series of 85 patients with thymoma seen at our teaching hospital. The aims of the study were (i) to evaluate both their frequency and occurrence in relation to the tumor course; (ii) to determine whether patients with AD other than MG have distinct epidemiological, clinical, and/or pathological features; and (iii) to determine risk factors for the occurrence of AD after thymectomy.

2. Methods

2.1. Study population

We retrospectively analyzed the clinical characteristics of patients who had a definitive pathologic diagnosis of thymoma, seen at three tertiary hospitals of the Hospices Civils de Lyon between 1/2005 and 12/2011. Patients were identified through the database of the three departments of Pathology of the Hospices Civils de Lyon. No approval from the University of Lyon Institutional Review Board was needed as per French policies; indeed, this retrospective study did neither cause any privacy violation nor lead to any intervention or treatment in the patients included. The authors observed a strict accordance to the Helsinki Declaration guidelines.

Clinical data were collected by a chart review while follow-up data were obtained by contacting general practitioners. Pathologic classification of thymoma was staged according to the World Health Organization classification: thymoma epithelial tumors comprising type A and B (developing respectively at the cortical and medullar of the thymus, or mixed for the type AB) and thymic carcinomas [3]. The clinical tumor stage was based on the modified Masaoka classification [16].

Complete remission of the AD was defined as the absence of any sign of disease activity, according to Osserman classification for MG, for example [17,18]; partial remission was defined as a significant improvement in disease signs with persistence of clinical and/or biological signs of disease activity.

All the general practitioners but 4 (lost to follow-up) were called in March 2015 to ensure up-to-date data analysis.

2.2. Literature review

We conducted a computer-assisted (PubMed, National Library of Medicine) search for publications up till June 2015. The articles included the discussed AD in thymoma patients, in general with the keywords “thymoma” and “autoimmunity” and by each AD represented in the study: MG, SLE, autoimmune cytopenia with PRCA, aplastic anemia (AA), autoimmune hemolytic anemia (AIHA), neurologic disorders other than MG with Isaac's syndrome, Morvan's syndrome and limbic encephalitis, polymyositis, Good's syndrome, autoimmune thyroiditis disorders with Grave's disease and Hashimoto thyroiditis, autoimmune hepatitis and cutaneous autoimmune disorders with paraneoplastic pemphigus and lichen planus. Our literature search was limited to the English and French language. The reference lists of all the articles were scanned for references not identified in the initial research. Only cases with well-documented clinical summaries and relevant information were included.

Download English Version:

https://daneshyari.com/en/article/3341500

Download Persian Version:

https://daneshyari.com/article/3341500

Daneshyari.com