Systemic vasculitis: how little we know about their societal and economic burden

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ABSTRACT

This article attempts to perform an evaluation of the state of the art of the economic and societal burden of systemic vasculitis (VAs). Due to the rarity of these diseases and their variable clinical picture, few data are available in the literature on their health economic issues, and only some papers have been published that marginally examine the problem.

Since VAs are severe conditions with a high medical and societal impact and determine high healthcare resource consumption, studies able to define societal, quality of life and economic burden of these pathologies are needed. Policy makers, private and public organisations involved in the care of VAs need data to programme future investment or make cost-effectiveness analysis for introducing new drugs or protocols.

Introduction

Systemic vasculitis (VAs) are an heterogeneous group of diseases, often characterised by a severe course. It may therefore be argued that, in addition to the impact on patients outcome and prognosis, these conditions, as many other rheumatic diseases, may have a high impact on the healthcare systems, the health care costs, and the society (1-3).

Aim of the present review is to examine the last decade literature studies on the issue of healthcare costs in VAs.

The review agrees the recommendations of the Centre for Reviews and Dissemination (4) and of the Cochrane Collaboration (5), thereby using an established rigorous and reproducible methodology. A protocol was developed to define review questions.

Methods

Published studies in English were searched using the main electronic database, PubMed MEDLINE. The search was performed for the period January 2002–September 19, 2012. The search strategy is as follows: ((“economics”[Subheading] OR “economics”[All Fields] OR “cost”[All Fields] OR “costs and cost analysis”[MeSH Terms] OR (“costs”[All Fields] AND “cost”[All Fields] AND “analysis”[All Fields]) OR “costs and cost analysis”[All Fields]) AND (“vasculitis”[MeSH Terms] OR “vasculitis”[All Fields])) AND ((“2002/01/01”[PDAT]: “2012/09/19”[PDAT]) AND “humans” [MeSH Terms] AND English[lang] AND “adult”[MeSH Terms]).

The publications were assessed for inclusion by a 3-step process: i. titles and abstracts of all identified studies were assessed by one reviewer and checked by a second reviewer; ii. full texts of relevant articles were then obtained and inclusion criteria applied independently by two reviewers. Possible discords between reviewers were resolved by consensus; iii. data were extracted by one reviewer and then checked by a second reviewer.

Inclusion criteria

In the study protocol the reviewers selected publications from the mentioned database as follows:

Period: Jan. 2002–Sep. 19, 2012
Language: English
Studies: all articles related to economic analysis
Patients: adult ≥18
Outcomes: direct costs, indirect costs

Exclusion criteria

Not published in English and all publications before 2002, conferences proceedings, case reports, reviews, systematic reviews, letters and commentaries were excluded.

Results

Up to September 19 2012, 35 articles were extracted, but only 3 papers were considered relevant to this review (Fig. 1) (6-8).

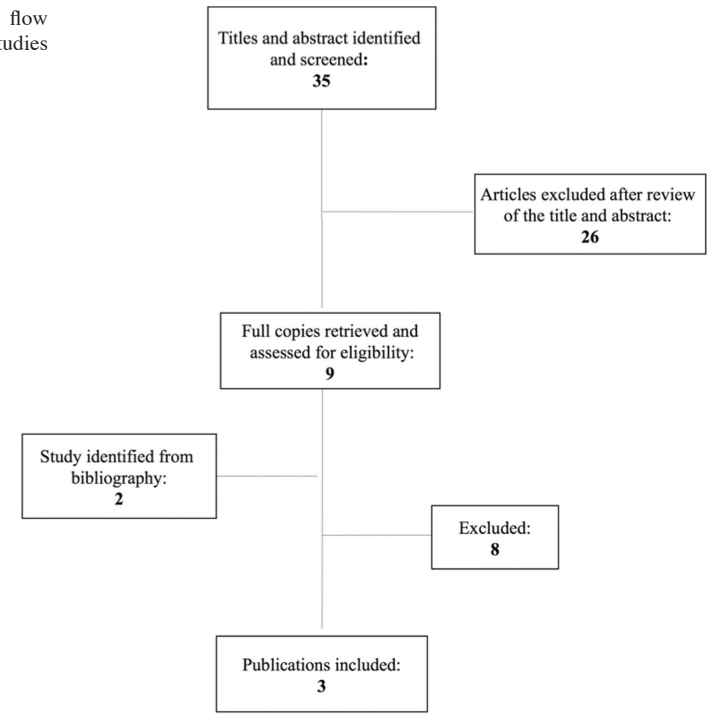

Fig. 1. PRISMA flow diagram for cost studies in vasculitis

Excluded articles had no indications of societal and/or economic impacts or considered only the impact of some therapies. Based on these results, a systematic literature review was not possible and therefore data are summarised below as a descriptive review.

The societal and economic impact of VAs

Single studies have examined the societal impact of VAs. Reinhold-Keller *et al*. (8) registered a very high frequency of hospitalisation and visits for Wegener's granulomatosis (WG) patients. More than the half of WG patients had been hospitalised in the year before the study. In extreme cases these authors registered one visit a week.

As far as the employment status was concerned, 27% of patients were unemployed because of the disease, while employed patients reported a mean of 14 workdays lost per year because of WG.

In this study, having WG diagnosis and being unemployed was associated with reduction of social and physical functioning.

Few studies specifically addressing costs in VAs are available. Krulichova *et al*. (6) estimate the amount of resources used and direct medical costs associated to active *versus* inactive Takayasu's arteritis (TA) in Italy, in a multicentre and prospective study.

The study reports €5054.3 *vs*. €1328.4 of mean annual costs per TA active and TA inactive patients, respectively. These costs are mainly generated by drug therapy (22.0% of the total costs) and hospitalisation (44.8% of total costs), measured using Diagnosis Related Groups (DRGs).

The direct and indirect costs associated to Behçet's syndrome (BS) are reported by Sut *et al*. (7). The study includes 119 Turkish BS patients divided into four groups based on the presence of ocular, vascular, neurological and mucocutaneous-joint involvement.

Direct annual per patient costs ($2203 on average) fall in a interval with a left-bound of $973 for mucocutaneous-joint patients' subgroup and a right-bound of $2727 for the ocular patients' subgroup.

Indirect annual per patient costs are $1023, on average. Highest costs have been registered within the neurological subgroup ($2190) and the lowest cost within the mucocutaneous-joint patients' subgroup.

As a result, the study reports $3226 of total annual per patient BS related costs with $5005 for the neurological subgroup, $1280, $679 and $207 within the ocular, vascular and mucocutaneous-joint subgroups, respectively. These results should be treated with caution because of the very high standard deviations reported.

Discussion and conclusions

The paucity of studies on direct and indirect costs of VAs made a systematic review not possible.

The few studies assessing direct costs suggest that VAs determine high costs related with their severity, the need for frequent hospitalisation and costly procedures.

Contrarily to other rheumatic diseases as far as indirect costs are concerned, although many data are available in the literature on outcomes and quality of life in these conditions (8-16), no detailed analysis are available on the consequences of a poor quality of life on employment, salaries and productivity.

The lack of a consolidated literature on this theme is a good opportunity to design and conduct *ad hoc* studies – also defining the criteria and the issues that must be included in the analysis – in order to obtain statistically significant and cross countries comparable results useful also for deriving health policy implications (17, 18).

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