
Socioeconomic consequences of ankylosing spondylitis

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ABSTRACT

Since ankylosing spondylitis usually starts at an early age, the socioeconomic impact of the disease on the patient and on society can be important. Several studies have reported substantial work disability, but there is considerable variation among countries. While withdrawal from work is associated with an older age at diagnosis and manual jobs or a lower educational level, sick leave in those with a paying job is associated with disease activity and physical functioning. The yearly mean total (direct and productivity) costs of AS are US\$6,720 per patient in the USA and Euro9,462 per patient in Europe when the human capital approach is used to calculate the productivity costs. A cost-effectiveness study on spa-therapy in AS shows that the clinical benefits of (expensive) treatments can be reached within acceptable costs.

Introduction

Within outcome research in ankylosing spondylitis (AS), the socioeconomic consequences of the disease have received limited attention. It is possible that its formerly reported low prevalence (1), its assumed good clinical outcome and the absence of effective treatments contributed to this restricted interest. The higher prevalence figures in more recent studies (2, 3), the awareness that the impact of AS on pain and function is comparable to that in rheumatoid arthritis (4) and the development of effective but expensive drugs (5,6) has resulted in increasing attention to the economic consequences. Socioeconomic impact refers to the effect of the illness on the ability to perform paid or unpaid work and on the effect on health resource utilization. Such consequences can be presented by describing in natural units the ability to work or the health resource use, but can also be valued in monetary terms resulting in an economic analysis (cost-of-illness). Importantly, the costs of management and consequences of a

disease should always be related to the health status and quality of life of the patient and ultimately the incremental cost-effectiveness and cost-utility analyses will be of interest for decision makers but also for clinicians and patients themselves.

Labour force participation in ankylosing spondylitis

A systematic review of the literature, completed with more recent publications, revealed 24 full papers and two abstracts addressing labour force participation in 18 different groups of patients suffering from AS (4, 7-14). In half of the studies employment was reported to be lower than 70% and in half of the studies work disability was reported to be higher than 20%. These figures were difficult to interpret especially because data were not adjusted for age and gender and because reference data of a comparison population were usually not provided.

One study among Dutch AS patients showed that the unadjusted LFP was 3% lower and work disability 27% higher when compared to the general population. However, after adjusting data for age and gender it was found that employment was 11% (OR: 0.82; 95% CI: 0.78-0.87) lower and work disability 15% (OR: 3.10; 95% CI: 2.77-3.42) higher than expected in the general Dutch population (11). It is likely that in studies presenting unadjusted figures, employment among patients is overestimated. Withdrawal from work in AS patients with a paid job was high in Mexico (3% per year) (15), intermediate in Europe (36% after 20 years in a French study and 5%, 21% and 31% after 1, 10 and 20 years respectively in a Dutch study) (8, 16) and low in the USA (5% after 5 years and 10% after 10 and 20 years respectively) (14). The Dutch study was able to calculate that age and gender adjusted withdrawal among patients was 3.1 times (95%CI: 2.6-3.7) compared to the general population (8, 9).

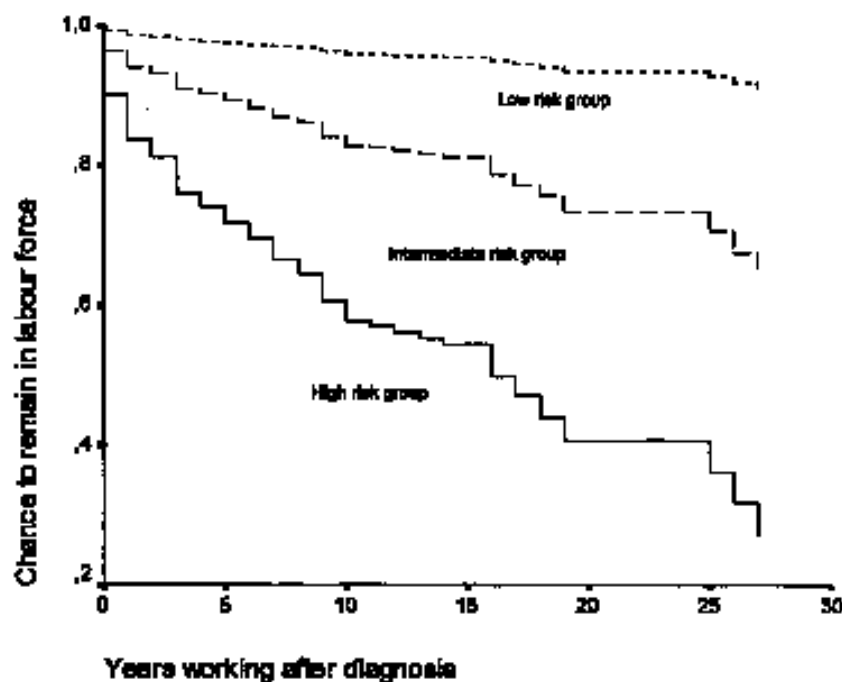


Fig. 1. Kaplan-Meier survival curve of the chance to remain in the labour force after a diagnosis of ankylosing spondylitis in three risk groups [Low risk group: no unfavourable coping strategies; Intermediate risk group: coping strategy characterised by limiting OR adapting activities; High risk group: coping strategy characterised by limiting and adapting activities].

In studies examining determinants of withdrawal from work it was shown that age (15), age at diagnosis (8,14), manual professions or low educational level (8, 14, 16) and the presence of comorbidity (14) were determinants of withdrawal. The Dutch study considered the effect of coping on withdrawal from labour force and a striking effect was observed. Coping strategies characterised by limiting or adapting activities were strongly associated with withdrawal (Fig. 1) (8). Interestingly, in a European tri-nation study among 210 Dutch, French and Belgian AS patients, it was shown that after adjusting for demographic and clinical confounders Dutch AS patients had a 3.82 times (95%CI: 1.33 – 11.0) higher risk to be work disabled compared to French or Belgian patients (10). The influence of the social security system or the economic status of the country on labour force participation was also seen in a German study among 8,776 patients with AS and 52,444 patients with RA. Patients living in the formerly Eastern states had a 2.38 increased risk to be work disabled compared to patients living in the formerly Western states. In

this study, the adjusted risk of work disability was 1.57 times higher among the RA patients compared to AS patients (4).

Sick leave in those with a paying job did not receive much attention as an outcome measure (7). This is surprising since sick leave is often the forerunner of (permanent) work disability. In the European two-year follow-up study it was shown that 37% of patients reported sick leave because of AS in the first year of the study and 45% in the second year (10). The average annual AS related sick leave was 13 days per patient and AS unrelated sick leave was 2.5 days per patient. In all countries the total sick leave was higher than the national sick leave. The length of sick leave was predicted by presence of inflammatory bowel disease (HR: 4.16; 95%CI: 1.22 – 14.29), higher disease activity (BASDAI) (HR: 1.34; 95%CI: 1.02 – 1.28) and living in The Netherlands compared to France or Belgium (HR: 1.54; 95%CI: 1.03 – 2.33).

Cost-of-illness in ankylosing spondylitis

One cost-of-illness (COI) study among

AS patients in the US (n = 241) was published as a full paper (17) and a COI study among European patients (n = 210) was published as a full paper for the productivity costs (10) and as an abstract for the direct costs (18). Both studies were prospective, considered the AS related health resource use and calculated direct (health care and non-health care) costs as well as productivity costs. The US study used mainly tariffs to evaluate resource utilisation, while for the European study true prices could be used. In the US cohort participants had a higher educational level and patients with IBD were excluded. When using the human capital approach (including loss of productivity because of sick leave and work disability until the age of retirement) the total COI in the US were US\$ 6,720 (median 1,495) per patient per year compared to Euro 9,462 (median Euro 2,894) per patient per year. At the time this supplement was written the exchange rate of both currencies varied considerably from week to week but was about one US\$ for one Euro.

The productivity cost accounted for 74% and 72% of the total costs in the US and Europe, respectively. Some health economists state that the human capital approach overestimates the productivity costs and advise to use the friction costs which takes only into account the production loss during the period of sick leave that the production is not yet replaced by another person (19). Calculated this way, total COI in Europe would be Euro 3,116 (median 1,490) per patient per year and would account for only 15% of the total costs. The direct (health care and non health care) costs were US\$ 1,775 (median 1,113) compared to Euro 2,640 (median 1,242) in US and Europe respectively. The main cost drivers for the direct costs in the US were the costs of drugs (42%) followed by costs of inpatient care (16%), while in the European study the main cost drivers were inpatient care (27%) followed by costs of drugs, physiotherapy and informal home care (each 13%). When compared to the COI in RA, as reported in a literature review (20), the mean total costs as reported in AS fall in the range

of costs reported for RA (Euro 2,841 to 10,602 per patient per year using 1998 consumer price index). Determinants of high costs of AS (>US\$10,000) in the US study were the amount of stiffness (OR 1.03; 95%CI: 1.00-1.05) and physical function (HAQ-S) (OR: 3.00; 95%CI: 1.49 – 6.03). The European study used a Cox regression analyses to assess risk factors for costs and identified higher age (HR: 1.12; 95%CI: 1.01 – 1.03), presence of peripheral arthritis (HR: 1.59; 95%CI: 1.23 – 2.27) and worse physical function (BASFI) (HR: 1.33; 95%CI: 1.25 – 1.45) as determinants of costs.

Full economic evaluations in ankylosing spondylitis

Only two cost-effectiveness studies are published in the literature (21, 22). Both were incremental cost-effectiveness analyses from societal perspective alongside a clinical trial. One RCT compared supervised group physical therapy with unsupervised exercises at home in 144 patients with ankylosing spondylitis (AS) during nine months (21). At the time of that study the present core set of outcome measures in AS for physical therapy were not yet defined (23, 24). At nine months the mean effects of group physical therapy and home exercises were, respectively, +0.9 cm (16%) and +0.5 cm (9%) for mobility ($p < 0.01$), +7 watts (4%) and -2 watts (-1%) for fitness ($p = 0.05$), and +1.7 (34%) and +0.3 (6%) for global health (VAS) ($p < 0.01$). Only direct costs were considered. It was estimated that the incremental costs of group physical exercise was Euro 400 (converted to 2000 Euros using the consumer price index) for a 7% increase in spinal mobility, 27% increase in aerobic fitness and 28% increase in quality of life. A second incremental cost-effectiveness analyses was also a piggy-back trial alongside an RCT comparing the effect of three weeks spa-exercise treatment at a spa-resort in Austria (group 1) with three weeks spa-treatment in The Netherlands (home country) (group 2) or usual care at home including group physical exercise (group 3) (22). After nine months, the cumulative gain in physical function (BASFI) for group 1 com-

pared to group 3 was 1.0 and for group 2 compared to group 1, the gain was 0.6 (scale 0-10). The cumulative gain in utility (EuroQol-5D) for group 1 compared to group 3 was 0.17 (scale 0-1) and for group 2 compared to group 3 the gain was 0.08. Direct (healthcare and non healthcare) and productivity costs were considered. The price of the spa-treatment in Austria was Euro 1,739 while the price of the spa-treatment in The Netherlands was Euro 1,515 per patient. During the study period the patients in the intervention groups used fewer NSAIDs and painkillers, had fewer visits to the physiotherapist and the patients in group one reported fewer days sick leave. The incremental cost-effectiveness ratio per unit effect gained in functional ability was Euro 1,269 (95% CI: 497 – 3,316) for spa-treatment in Austria and Euro 2,477 (95% CI: 601 – 12,089) for for spa-treatment in The Netherlands (home country) compared to controls. The incremental cost-utility ratio per unit effect gained in quality of life was Euro 7,465 (95% CI: 3,294 – 14,686) for group 1 and Euro 18,575 (95%CI: 3,678 – 114,257) for group 2 compared to controls. In the Dutch society these results are considered acceptable cost-effectiveness values.

Conclusion

Sick leave, work disability and withdrawal from work are increased among patients with AS when compared to subjects from the general population. There are substantial differences in working status among the countries. Older age at onset of disease, manual jobs or lower educational level and coping characterised by limiting or adapting activities are associated with withdrawal from work while disease activity is a determinant of sick leave. The costs of AS in USA and Europe fall within the range of costs as reported in RA. Worse physical function is an important predictor of total costs. From two CEA trials comparing group physical exercise or spa-exercise treatment with usual care, we learn that the benefits of (expensive) interventions can be reached at acceptable costs-effectiveness.

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