Productivity Losses and Costs in the Less-Common Systemic Autoimmune Rheumatic Diseases

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Abstract (153 words)

Purpose of Review:

We synthesized the literature on productivity losses and costs in the less-common systemic autoimmune rheumatic diseases: Sjogren's syndrome (SjS), systemic sclerosis (SSc), poly/dermatomyositis (PM/DM), and systemic vasculitides (SV).

Recent Findings:

Of 29 studies located, 12 were published 2012-onwards (SSc=6, SjS=2, PM/DM=2, SV=2). In these, 25% of PM/DM, and 21%-26% of SV, were work disabled, 22% of SSc stopped work within three years of diagnosis, and annual costs of absenteeism in SSc averaged $12,024 2017 USD. Very few studies reported on costs, presenteeism (working at reduced levels), or unpaid productivity loss. Across multiple SARDs, major drivers of lost productivity were generalised items like pain, depression, and fatigue, rather than disease-specific factors.

Summary:

Evidence suggests that work disability is common in SSc and strikes quickly. However in SSc and other SARDs, more comprehensive estimates are needed, which include absenteeism and presenteeism from paid and unpaid work, costs, and drivers of productivity loss.

Introduction

Systemic autoimmune rheumatic diseases (SARDs) are a group of chronic arthritides including systemic lupus erythematosus (SLE), Sjogren's syndrome (SjS), systemic sclerosis/ scleroderma (SSc), poly- and dermatomyositis (PM/DM), and forms of adult systemic vasculitides. Though distinct disorders, SARDs are often studied together due to shared pathogenesis, manifestations, and treatments.

Immune dysregulation in SARDs results in systemic inflammation, organ damage, and a high morbidity burden that is compounded by the adverse effects of immune-modulating and immunosuppressive therapies. Altogether the physical, psychological, and neurocognitive manifestations of SARDs can lead to high levels of healthcare resource use and costs[1–6] and reduced health-related quality-of-life[7–12], and limit participation in paid employment[13,14], and family[15–17] and other meaningful activities[18,19]. These effects can strike early, with 26% of working-age SjS[20], and 28% of SSc[21], unable to work for pay within two years of diagnosis. In earlier reports from Canada, the lost productivity costs of SLE and SSc exceeded the direct medical costs (i.e. costs for hospitalisations, physician visits, and medications) by two- to three-fold[22–24].

Productivity losses, which encompass time lost from paid and unpaid work activities, take many forms including work disability (leaving the paid workforce work due to health), absenteeism (work time missed due to health), and 'presenteeism' (working, but at a reduced level/efficiency). Estimates of lost productivity are useful in informing health policy, allocating scarce resources, and setting research and treatment priorities. Moreover, knowledge of the

drivers and predictors of productivity loss can inform efforts to help reduce (or even prevent) these losses.

SLE is the most common SARD and several reviews[13,25–29] are available on the productivity burden of this disease. But while recent reports show other SARDs, such as SSc, PM/DM, and systemic vasculitides (SV), being associated with high healthcare costs[2,3,5,6], their productivity burden remains largely unknown. A 2012 review on the economic burden of SV[30] profiled just two productivity studies, and while two systematic reviews are available on employment in SSc[14,31], they do not include studies published after 2012. Here, we review the existing literature on productivity losses in these 'lesser-known' SARDs (SSc, Sjogren's, poly/dermatomyositis, and systemic vasculitides) to summarize what is known about the extent and drivers of their productivity burden, and identify gaps and directions for future research.

How Are Productivity Losses Assessed?

While an extensive discussion of productivity and health is beyond the scope of this review, we begin with a brief overview of the components of productivity loss and how they are measured and valued. Readers wishing more information are encouraged to consult the following resources[32–35].

A compromised health state can affect one's performance of both paid and unpaid work, with the specific types of unpaid work (i.e. housework, yard work, childrearing, studying, volunteering) varying across studies. Lost productivity can be measured as a categorical outcome (employed: yes/no) or continuously (*absenteeism* and *presenteeism*), in units of time

(i.e. hours or days of presenteeism, absenteeism, or long-term disability), money, or both. Estimates of productivity costs (sometimes called indirect costs) are produced by assigning a monetary value to the time estimates (i.e. multiplying hours/days by an hourly/daily wage). Although there is now a movement away from using the terms *direct* and *indirect*[33], direct costs refer to costs for the provision of goods or services, such as laboratory tests or medications. Productivity costs are considered indirect since costs are incurred (i.e. a business's production decreases when an employee is not working at full capacity) but not paid out to any party. While productivity costs are not always considered by healthcare payers in decision-making, they are relevant from a societal perspective, and should be assessed and reported when possible[35].

Estimates of lost productivity costs will vary depending upon the methods used to value work. If time losses from unpaid work are valued using the average wages of individuals hired to do the same work, such as housecleaners or childcare workers (the replacement cost method[33]), costs will usually be lower than when using the same wages one would receive in their paid job (the opportunity cost method[33]). Bowman *et al.*'s[36] 2010 estimates of the lost productivity costs of Sjogren's in the United Kingdom (UK) illustrate how cost estimates are influenced by the components included, and how they're valued. They considered time patients missed from paid and unpaid work, and time helpers missed from paid work when accompanying the patient to medical appointments. Employed patients were asked about the actual number of sick days taken, additional hours they would regularly work in the absence of health problems, and additional weeks of work missed per-year due to illness. Time losses from unpaid work were valued using both replacement and opportunity costs. With these different measures of patients' and carers' paid work loss, and patients' unpaid work loss, they produced

six estimates ranging from $12,730 (converted to 2017 USD), which included sick days from paid work, and valued unpaid work losses with the replacement wage, to $22,390, which included all three aspects of paid work loss, and used sex- and age-matched average wages to value unpaid work losses.

The measurement and valuation of productivity losses is subject to equity concerns that are especially relevant in SARDs, as they predominantly affect females. Some SARDs, like SLE, strike mainly between the ages of 20 and 50, patients' peak career and childrearing years. Others, like Sjogren's and giant cell arteritis, affect mainly those over 50, some of whom may already have retired. Estimates of paid work losses alone will exclude the time contributions of homemakers, students, and those retired for health or other reasons. Valuing productivity losses with age- and sex-specific wages can also undervalue the time of females (whose wages are typically lower), and those in lower-paying occupations. The implications of these choices are illustrated in a well-cited paper by Clarke *et al.*[37], who assessed the annual productivity costs of females with SLE under five scenarios that varied the type of work considered (paid-only, or paid and unpaid), wages (overall or sex-specific), and valuation of unpaid time (replacement or opportunity costs). Estimates ranged from $9,776 (2017 USD), for 12-month losses from paid work, to $21,120, when including time lost from paid and unpaid work, and using males' average wages to value unpaid productivity loss.

Prior Work

Numerous studies have been conducted on work disability (WD) and productivity losses and costs in SLE, and these have been summarised and critiqued in several reviews[13,25–29].

In a systematic review and meta-analysis of 26 studies[13], 46% (95% CI: 40%-52%) of SLE were employed while 34% (24%-44%) were work disabled (WD). Patient factors associated with WD included lower education and socioeconomic status, decreased physical functioning, higher disease activity, and higher levels of pain, fatigue, and anxiety, while job/workplace factors included greater physical demands and less control. A systematic review on costs of SLE[25] found that poor physical or mental health, less social support, higher educational attainment, and high disease activity were associated with higher lost productivity costs

Therefore, while gaps remain in our understanding of lost productivity in SLE, we decided to focus this review on the other SARDs (SSc, Sjogren's, poly/dermatomyositis, and SV), where far less is known about their productivity burden. We will briefly review the earlier literature (published before 2012), before detailing and critiquing the most recent reports. Our review is restricted to studies published in English (thus excluding at least two articles[38,39]), and reporting quantitative data on employment/work disability rates, absenteeism, presenteeism, or costs. All reported costs were converted to 2017 USD.

Systemic sclerosis (SSc)

SSc was discussed in the majority (11/17) of studies (Table 1). Lost productivity costs, reported in three[22,40,41], ranged from $8,592 in Hungary, to $13,796 in the United States (US), and $14,584 in Canada. Forty percent of costs in the Canadian cohort were for time lost from paid work (43% of costs for the diffuse subset (dcSSc) and 36% for the limited (lcSSc)). Nine studies assessed WD, with prevalence ranging from 19%[41] to 61%[42] (Table 1). In one[43], WD was significantly more frequent in SSc than RA (56% vs. 35%, p=0.01), while in a

tenth study[44], employment rates were significantly lower in SSc than the general population (standardised employment ratio=0.77). Determinants of WD and productivity varied, but included disease duration[21,43,45], subtype[21,43], hand function[45–47], fatigue[21,46–48], pain[21,47], education[48], social support[48], and self-rated health[42,46]. Nearly all were cross-sectional studies, but in the one longitudinal investigation[48], lung involvement and fatigue at enrollment, and non-Caucasian ethnicity, were significant predictors for *becoming* WD.

Sjogren's syndrome (SjS)

The lost productivity costs of SjS were reported in the Bowman *et al.*[36] paper described previously, while WD was reported in two others. In a US study[49], 12% were WD (versus none in the sex and age-matched comparison group), and WD individuals had higher levels of pain, depression, and cognitive dysfunction than non-WD. A Dutch report[50] found that SjS were less likely to be employed full-time (10% vs. 24%) or at all (51% vs. 83%), compared to the general population, and employed SjS worked fewer hours per-week than non-SjS (22 vs. 27). Higher education was associated with being employed.

Systemic vasculitides

The earlier studies reported on just one condition, Granulomatosus with polyangiitis (GPA), formerly known as Wegener's disease. In related studies of GPA patients in the US and Netherlands, 77% of employed GPA in the US cohort (80% in the Netherlands[51]) reported taking more than six consecutive weeks of sick leave, with 57%[52]/53%[51] US/Dutch taking more than six consecutive months of leave. More than half had to modify their jobs or take total

disability, with 51%[52]/41%[51] reducing work hours, 37% changing duties[52], 17%[52]/5%[51] resigning, and 31% of the US cohort (27% of the Dutch[53]) receiving disability benefits. Reinhold-Keller *et al.*[54] studied a German cohort of prevalent GPA who were aged ≤ 40 years at diagnosis. Sixteen of 60 individuals were WD, and far more females than males (78% vs. 29%) had become unemployed since GPA diagnosis, though not all work loss was due to GPA. Finally, in another German analysis[44], the standardised employment ratio for GPA (0.76) was the lowest among all six rheumatic diseases studied.

Recent Research (2012-present)

Turning our attention to the most recent work in the field, we located 12 studies published in the past five years (2012-onwards), whose characteristics are in Table 2. Eight were from Europe[20,55–61], one Australia[62], two the US[1,63], and one was from Canada[64]. Six reported on SSc[55,57–59,62,63], two on SV[61,64], two on PM/DM[1,56], and two on SjS[20,60]. As the earlier estimates focussed mainly on SSc, it was encouraging to see more studies emerge on other SARDs. However, some gaps remain, with few studies reporting on presenteeism[63,64], patients' unpaid work loss[63], or costs[57,59,63]. Moreover, heterogeneity in the definitions and measures of productivity loss (i.e. employment yes/no, work disability for any health reason versus the SARD specifically, days versus hours of presenteeism), and methods used to monetise these losses, limits the comparability of estimates across studies.

Systemic sclerosis (SSc)

Three studies in SSc reported on productivity costs. In the first[57], which had estimates from seven European countries, carers' time losses from paid work were valued using country-specific wages for professional caregivers, and absenteeism for patients was valued using country-specific (and, in at least one country[65], sex-specific) average wages. Unlike most studies, which recruited through tertiary clinics, this one recruited through patient organisations. That should have reduced selection bias, though in France, where participants were recruited exclusively online, the percentage of the cohort with a university degree was 2.2 to 4.1-times higher than the general population[65]. Mean costs for informal caregiving ranged from $0 in Sweden to $831 in Germany and $6,554 in Spain. Patients' lost productivity costs ranged from $2,380 in Italy to $20,210 in the UK, with early retirement tending to account for the majority of costs. The authors could not fully explain this wide disparity in costs between countries, but suggested that differences in wages and labour and social legislation, as well differing proportions of SSc with limited versus diffuse disease, may have contributed. The small number of respondents in some countries (i.e. 24 SSc patients in the UK and 23 in Sweden; 0 carers in Sweden and only 3 in each of the UK, Hungary, and Germany) may also have skewed the estimates.

In the second study[63], members of the UCLA Scleroderma Quality of Life cohort were asked about days of absenteeism (work days missed) and presenteeism (days where work productivity was reduced by $\geq$ 50%) from both paid and unpaid work. Twenty-four percent of the cohort were WD due to SSc; factors significantly associated with WD included higher disability (measured by the Health Assessment Questionnaire Disability Index (HAQ-DI)) and lower education. Among those in the paid workforce, those with lcSSc had significantly more

days of absenteeism and presenteeism than dcSSc. Lower education and poorer overall health were associated with productivity loss from paid work, while having lcSSc or worse HAQ-DI scores were associated with productivity loss from unpaid work. Lost productivity costs (defined as lost earnings, which may not represent the societal costs) for those in paid work were valued using sex- and education-specific median weekly earnings, and averaged $1,002 per-month ($12,024 annually); those for WD individuals averaged $47,952 annually. Despite this being one of few studies reporting on presenteeism and unpaid productivity losses, costs were not computed for these components, perhaps because time loss was measured in days instead of hours. Moreover, as with the aforementioned European estimates, there are equity concerns associated with the use of sex-specific wages to value time loss in a disease that predominantly affects females.

In the third cost study[59], data from the Social Insurance Institution in Poland were used to assess the costs of sick days, and short- and long-term disability, from paid work. Depending on the method of valuation, mean per-person costs ranged from $3,876 to $12,677. Sick days accounted for 19% of costs, short-term disability 5%, and long-term disability 77%. While the national social insurance database made the estimates more generalisable, neither presenteeism, nor productivity loss from unpaid work, were captured in the database, so were not included in the costs.

Three other studies reported on WD in SSc, but not costs, with 40% of a working-age, clinic-based cohort in Belgium being WD[58] and 20% of an Australian clinic-based cohort not employed[62]. Although working-age participants in the former study were asked about several

health-related work transitions, the majority had stopped working entirely instead of reducing their hours or changing jobs. In the latter study, having dcSSc, a physical job, pulmonary hypertension, sicca symptoms, or digital amputation were all significantly associated with unemployment.

Finally, in the first of two longitudinal studies from Sweden (one on SSc and one on SjS), routinely-collected data were used to assess WD in a working-age cohort of newly-diagnosed SSc, and a reference group from the general population[55]. WD was assessed each month, continuously and categorically (any sick leave or disability days), using data from the Swedish Social Insurance Agency. Of the 25 SSc who had no WD at diagnosis, 25% had more than 90 days of WD during the first three years *after* diagnosis, and 22% were on full-time WD. Those with dcSSc averaged more days of WD, and were more likely to develop full WD. No disease-related factors correlated significantly with WD, but education level, number of years in the workplace, and sickness absence in the month prior to SSc diagnosis were correlated with cumulative WD days over the three years following diagnosis.

Sjogren's syndrome (SjS)

The second longitudinal study[20] employed a similar design, and sourced the same routinely-collected data, to study WD in a cohort of working-age SjS. Twenty-six percent of SjS had any WD at diagnosis, and this number rose to 37% at 12 months, and 41% at 24 months. Older age, being on sick leave or disability at the time of SjS diagnosis, and having fibromyalgia, all increased the odds for being WD at 24 months, though disease activity did not. Days of WD averaged 6.2 per-month at the time of diagnosis, 9.2 at 12 months, and 10.2 at 24 months. A key

feature of these two Swedish studies was the population-based data source, which minimised selection and recall bias, and enabled comparison between SSc/SjS and the general population (four non-SSc/SjD individuals for each SSc/SjD). However, data were not available on presenteeism or time loss from unpaid work, nor absences of less than 14 days, since these losses are not compensated by the Swedish Social Insurance Agency.

German investigators compared the productivity of working-age females with SjS attending one of four tertiary clinics, with a sample of their similarly-aged female friends without SjS[60]. Fifty-three percent of SjS were employed, as were 77.1% of non-SjS. Employed SjS reported more days of sick leave over the past six months than non-SjS (19.8 vs. 4.5), and 10.4% of SjS (vs. 1.5% of non-SjS) were on currently sick leave. In addition, retired SjS had stopped working earlier than retired non-SjS, at mean age of 47.9±8.5 vs. 57.5±4.5 years. Age, disease duration, functional capacity, and lack of stamina were all predictive of employment status, while pain and dryness were not. Though admirable that data were collected for a non-SjS comparison group, one concern is that 'friend controls' (used in an earlier SjS study[49] as well) may not be representative of the broader population.

Systemic vasculitides (SV)

Two recent studies have extended our knowledge of productivity loss in SV beyond GPA. In one, conducted at a tertiary rheumatology centre in Ontario, Canada[64], 43% of 51 working-age SV were WD, defined as not working (59%), took early retirement (14%), or working reduced hours (27%). WD participants had greater functional impairment (HAQ scores of 0.86 vs. 0.14), more pain, organ system involvement, and disease activity and damage, and

were less educated. Among those still employed, productivity loss due to health (measure of presenteeism) averaged 8.2%, and was correlated with HAQ and pain scores, but not age, disease duration, or levels of disease activity or damage.

In the second, Basu *et al.*[61] assessed factors associated with WD in a hospital-based, UK cohort of working-age individuals with ANCA-associated vasculitis. Twenty-six percent were WD (not working due to health), with fatigue, overweight, depression, and severe disease damage all increasing the odds of WD, while no other clinical factors (i.e. disease activity, subtype, therapies, ANCA status) did. Unfortunately, neither of these studies had data for a non-SV comparison group, and while the Canadian study only reported on WD that participants considered SV-related, this may be difficult to attribute.

Polymyositis/dermatomyositis (PM/DM)

In 2016, the first-known studies of productivity loss in PM/DM were published. One, reporting on a Swedish myositis registry[56], found 44% of working-age participants were working full-time, and 31% part-time, while 25% were on full-time sick leave. The second used a commercial healthcare claims database to estimate medically-related work loss (absenteeism and disability claims) for a cohort of employed PM/DM with disability insurance, and a non-PM/DM matched comparison group[1]. Though disability claims were captured directly from the database, no other productivity data were available, so estimates of medically-related absenteeism were determined from healthcare utilisation claims. An emergency room visit or day in hospital during the work-week counted as one full day of missed work, and outpatient medical visits counted as a half-day. They found that PM/DM averaged 2.0 more days of work

loss over a 12-month period than did the non-PM/DM, driven more by medically-related absenteeism.

While the administrative claims database allowed for a larger sample size than the registry study (n=611 versus 48), and facilitated access to data for a non-PM/DM comparison group, it did not allow for assessment of presenteeism or unpaid work loss. Furthermore, the use of healthcare encounters as a proxy for medically-related absenteeism was problematic, with the authors themselves acknowledging that they did not know whether utilisation actually occurred during individuals' working hours.

Discussion

In summarising the literature on productivity losses and costs in the lesser-known SARDs (SSc, Sjogren's, poly/dermatomyositis, systemic vasculitides), we located 29 studies, with 12 published over the past five years. Most focussed on SSc, and in these, SSc was associated with high rates of work disability and substantial productivity costs. However, there remain several gaps in the literature that are limiting our understanding of the incremental productivity losses and costs of SSc and other SARDs. The types of productivity losses that have been assessed, how they were monetised, and the study populations and data sources used, merit further discussion.

Completeness of Estimates

Although presenteeism is recognised as a major driver of productivity loss[32], accounting for 41% of losses in one study of arthritis[66], just three of 29 studies reported on presenteeism[45,63,64], and there were little data on productivity losses from unpaid activities[22,36,41,45,63]. Ideally, productivity studies should collect data on participation in paid work (work cessation and job changes), as well as absenteeism and presenteeism from paid and unpaid work, and monetise these losses in an equitable manner. Doing so would provide more complete estimates of the productivity burden, ones with greater comparability. The cost estimates discussed in this review (from seven studies[22,36,40,41,57,59,63]) varied widely, even after standardisation to 2017 US dollars. Though some of this variation can be attributed to differences in patient populations, and transnational differences in purchasing power, and labour or disability policies, there was considerable heterogeneity in which aspects of productivity loss were included, and how they were valued. For example, paid work losses were valued using

overall average wages/earnings in at least one study[40], and sex-specific (and sometimes age- or education-specific) wages in three[36,41,63]. Unpaid work losses were excluded from some studies altogether, while one did report time losses from paid and unpaid work, but only determined the costs of paid work loss[63]. Others determined the costs of paid and unpaid work losses, but applied the replacement wage to unpaid work losses[22,41], which are typically lower than the overall-average or sector-specific wages used to value paid work.

These practices limit comparability of estimates, and, more importantly, undervalue the economic contributions of those whose health has left them unable to participate in the paid workforce, as well as those not-employed for other reasons (i.e. homemakers, students, and retirees). Eliciting patients' unpaid work losses can also reveal the trade-offs some patients make between time spent on paid and unpaid work[32], and the resultant costs. Employment offers many benefits, and those with health impairments may continue in their paid work for personal and social rewards, to meet current financial needs, or remain eligible for pensions and insurance. But participation in paid work can leave them with limited time or physical capacity to complete household tasks, especially if their paid working tasks take longer to complete[32].

Data Sources

The cohorts in most studies in this review were recruited from academic/tertiary medical centres. It may be easier to contact these individuals and collect data from them on short-term work absences, presenteeism and unpaid work losses; information on potential drivers of productivity loss like disease activity, lung function, pain, and fatigue, are also more readily available. But one must consider that patients attending these clinics may not be representative

of others with SARDs: they may have more severe disease, or possibly be in a better socioeconomic position. An ideal source of data would build upon those used in the Swedish studies (which linked routinely-collected data for the general population with patients' clinical and sociodeomographic data), while also providing self-reported health status and productivity data for patients, and a sample of the general population. Such linked, population-based data from SARD and non-SARD individuals would eliminate the need for patients to attribute productivity losses to their SARD specifically, and provide policymakers with better estimates of the incremental productivity burden of these disorders.

Future Directions

An important finding from our review was that the main drivers of lost productivity are often not disease activity or other disease-specific items[20,47,58,60,61,64], but more generalised factors such as being overweight[61], pain[21,47,49], depression[49,61], cognitive dysfunction[49], and fatigue[21,46–48,60,61]. Most were cross-sectional studies of prevalent cohorts, so it's unclear whether factors like fatigue and depression contributed to WD, or developed afterwards (and perhaps even resulted from it). More investigations of incident cohorts would help address this. Still, higher baseline fatigue was predictive of WD in one of the longitudinal studies we located[48], and depression and cognitive function have been predictive of subsequent work loss in SLE[67].

Another unanswered question is how the productivity burden of these SARDs may have grown, lessened, or shifted over time, as has happened to a limited extent in rheumatoid arthritis

(RA)[68,69]. For example, while there was no net change in productivity costs between cohorts of patients with early RA diagnosed in different time periods (1996-1998 and 2006-2009)[69], the later cohort had fewer costs from sick leave than the early cohort, and more costs from disability pension. Many studies included in this review reported an inverse relationship between educational attainment and WD/productivity loss. With females attaining higher levels of education over the decades, it is possible that the productivity costs of SARDs have decreased alongside. While secular changes could not be evaluated from the current literature, this is a key area for ongoing research, with implications for patients and policymakers. In addition to controlling for changes in patient characteristics and treatment practices over time, such studies should collect data on all aspects of productivity loss, in order to capture secular shifts in costs (i.e. from absenteeism to presenteeism, or paid to unpaid time losses) that may otherwise go unrecognised.

Until now, research on individual SARDs has been limited by the relatively small number of people living with each disorder. However, SARDs have shared pathophysiology and manifestations, and now appear to share some drivers of productivity loss. Several educational, psychological, and exercise interventions have been effective at reducing depression and fatigue in SLE[70,71] and SjS[72]. Thus, while more work is needed, it is possible that productivity losses in SARDs could be mitigated (and perhaps prevented) by lowering levels of pain, fatigue and depressive symptoms. It is also promising to think that knowledge generated about reducing productivity losses in one SARD could be applied to another.

Conclusions

While the body of literature on productivity losses in SARDs has grown, there is a need (especially in poly/dermatomyositis and systemic vasculitides) for more comprehensive estimates, from incident cohorts, that include absenteeism and presenteeism from paid and unpaid work, calculate costs, and ideally, assess transitions to WD and drivers of these losses on a longitudinal basis. This knowledge would inform decision-making and priority setting, and potentially contribute to the development of strategies and interventions aimed at reducing the economic and societal burden of these lifelong diseases.

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Recent Important References

One of the first-known publications on productivity losses in PM/DM

- Regardt M, Welin Henriksson E, Sandqvist J, Lundberg IE, Schult M-L. Work ability in patients with polymyositis and dermatomyositis: An explorative and descriptive study. Work Read. Mass. 2015;53:265–77.

Recruited patients and their caregivers through patient organisations (instead of academic medical centres) and a rare diseases registry, and computed the productivity costs of SSc separately for seven European countries

- The BURQOL-RD Research Network, López-Bastida J, Linertová R, Oliva-Moreno J, Serrano-Aguilar P, Posada-de-la-Paz M, et al. Social/economic costs and health-related quality of life in patients with scleroderma in Europe. Eur. J. Health Econ. 2016;17:109–17.

Recent Very Important References

Linked data from a clinical registry, with population-based social insurance data for incident cases (SjS and SSc) and a sample of the general population, in order to assess work disability longitudinally from the time of SjS/SSc diagnosis

- Sandqvist G, Hesselstrand R, Petersson IF, Kristensen LE. Work Disability in Early Systemic Sclerosis: A Longitudinal Population-based Cohort Study. J. Rheumatol. 2015;42:1794–800.
- Mandl T, Jørgensen TS, Skougaard M, Olsson P, Kristensen L-E. Work Disability in Newly Diagnosed Patients with Primary Sjögren Syndrome. J. Rheumatol. 2017;44:209–15.

Table 1. Characteristics and Lost Productivity Data for Studies Published Through 2011

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Sjogren's syndrome (SjS)																
Bowman, 2010[36]	2003-2005	United Kingdom	cross-sectional	self-report questionnaire	clinic-based	clinic-based sample of patients with RA; patients without SjS or RA recruited from a local general practitioner	84 pSjS, 87 RA, and 96 non-SjS	84 (100%)	60 (11)	7 (7)	26/84=31% of SjS and 68/96=71% of non-SjS employed (mean 26.7 and 33.3 hours worked per week); 15% of SjS and 49% of non-SjS employed full-time (mean 35.0 and 39.0 hours worked per week)	annual mean hours of missed work: 35.8 (95% CI: -3.3 - 74.9) for SjS and 22.5 (6.9 - 38.1) for non-SjS; annual mean hours of missed work for carers: 10.5 (2.3 – 18.7) for SjS and 1.6 (-0.3 – 3.4) for non-SjS	-	annual mean hours of household work loss: 146.3 (70.2 – 222.4) for SjS and 35.1 (-3.7 – 74) for non-SjS;	annual time losses from paid work: £6,155 to £11,612 for SjS and £540 to £2,937 for non-SjS; annual time losses from household work: £1,376 to £1,745 for SjS and £330 to £423 for non-SjS; annual time losses from paid work for carers: £146 for SjS and £22 for non-SjS [2008 British pounds]	annual time losses from paid work: $10,193 to $19,230 for SjS and $894 to $4,864 for non-SjS; annual time losses from household work: $2,279 to $2,890 for SjS and $547 to $701 for non-SjS; annual time losses from paid work for carers: $242 for SjS and $36 for non-SjS
Segal, 2009[49]	2007	United States	cross-sectional	self-report questionnaire	clinic-based	unrelated friends (same sex and age) of patient members of the Sjogren's Syndrome Foundation	277 pSjS and 606 non-SjS	90%	62 (12.6)	9.0 (8.4)	12% of SjS and 0% of non-SjS not employed due to disability	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Meijer, 2009[50]	n/a?	Netherlands	cross-sectional	self-report questionnaire	clinic-based	age- and sex-specific Dutch population data	195: pSjS=154 sSjS=41; 135 of working age: pSjS=109 and sSjS=26	179 (92%)	55.5 (15.0)	9.7 (8.8)	63/135=47% of SjS, 49/109=45% of pSjS and 14/26=54% of sSjS (vs. 2% of general population), receiving disability benefits; 69/135=51% were employed, 58/109=53% of pSjS and 11/26=42% of sSjS (vs. 83% of non-SjS)	15.6±39 days of sick leave per year, 14.7±37.8 for pSjS and 22.3±50.0 for sSjS	-	-	-	-
Systemic Sclerosis (SSc)																
Bérezné **, 2011**[45]	2008-2009	France	cross-sectional	self-report questionnaire	clinic-based and French SSc patient association	none	189	164 (87%)	54.1 (13.3)	9.3 (8.4)	36/113=32% of working-age receiving full disability pension	-	mean SSc-related decrease in productivity of 3.4±3.8 over the past month (range 0-10)	mean hours of SSc-related household help per-month: paid=4.0 ±13.5 (8.0±21.5 for those with DU and 2.0±6.3 for those without DU); unpaid=9.0 ±27 (18.7±40.8 for those with DU and 4.0±15 for those without DU)	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Bernatsky, 2009[22]	2007?	Canada	cross-sectional	self-report questionnaire	clinic-based	none	457: lcSSc=272 dcSSc=185	401 (88%)	55.1 (12.1)	10.5 (8.6)	-	-	-	-	$13,415 overall ($5,345 from paid work and $8,070 from unpaid); $11,277 for lcSSc ($4,101 from paid work and $7,176 from unpaid); $16,416 for dcSSc ($7,092 from paid work and $9,324 from unpaid) [2007 CDN]	$14,584 overall ($5,811 from paid work and $8,773 from unpaid); $12,260 for lcSSc ($4,458 from paid work and $7,801 from unpaid); $17,846 for dcSSc ($7,710 from paid work and $10,136 from unpaid)
Hudson, 2009[21]	2004-2008	Canada	cross-sectional	self-report questionnaire	clinic-based	none	365	83%	WD: 50.2 (8.2); non-WD: 48.4 (9.4)	WD: 11.0 (8.6); non-WD: 9.0 (7.7)	133/365=36% of working-age participants; 133/643=21% of all participants	-	-	-	-	-
Mau, 2005[44]	1993-2001	Germany	cross-sectional	self-report questionnaire	clinic-based	German population data	802	667 (83%)	47 (10)	n/a?	SER=0.77 (0.85 for males and 0.75 for females); adjusted RR for employment (vs. RA reference group) of 0.98 (disease duration ≤ 5 years) and 1.03 (disease duration > 10 years)	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Minier, 2010[40]	2006?	Hungary	cross-sectional	self-report questionnaire	clinic-based	clinic-based samples of patients with RA and PsA	80: 60 lcSSc and 20 dcSSc	72 (90%)	57.4 (9.6)	6.2 (6.6)	39/80=49% receiving disability benefits (vs. 35% of RA and 25% of PsA)	-	-	-	mean annual informal care: €246 overall, €197 for lcSSc and €393 for dcSSc mean annual disability pension: €5,305 overall, €5,025 for lcSSc and €6,142 for dcSSc mean annual sick leave: €85 overall, €111 for lcSSc and €8 for dcSSc total lost productivity costs (sick leave and pension): €5390 overall, €5134 for lcSSc and €6150 for dcSSc [2006 Euros]	mean annual informal care: $392 overall, $314 for lcSSc and $626 for dcSSc mean annual disability pension: $8,456 overall, $8,010 for lcSSc and $9,790 for dcSSc mean annual sick leave: $136 overall, $177 for lcSSc and $13 for dcSSc total lost productivity costs: $8,592 overall, $8,187 for lcSSc and $9,803 for dcSSc
Nguyen, 2010[42]	2007	France	cross-sectional	self-report questionnaire	clinic-based and French SSc patient association	none	87	72 (83%)	48.6 (8.5)	8.1 (6.4)	53/87=61% on full-time sick leave; 31/87=36% receiving disability benefits	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Ouimet, 2008[43]	2002-2003	Canada	cross-sectional	self-report questionnaire	clinic-based	clinic-based sample of patients with RA	61: 35 lcSSc and 26 dcSSc	52 (85%)	52.0 (1.2)	11.0 (1.2)	34/61=56% overall (95% CI: 43%-68%), vs. 36/104=35% (26%-44%) of RA 17/26=65% of dcSSc, 17/35=49% of lcSSc	-	-	-	-	-
Sandqvist, 2008[46]	n/a?	Sweden	cross-sectional	patient interview	clinic-based?	none	44 lcSSc	44 (100%)	median 52 (range 24-60)	median 8 (range 2-44)	23/44=52% on full or partial sick leave: 15/44=34% partial and 8/44=18% full sick leave or disability	-	-	-	-	-
Sandqvist, 2010[47]	2008	Sweden	cross-sectional	self-report questionnaire	clinic-based?	none	57: 47 lcSSc and 10 dcSSc	53 (93%)	median 58 (IQR 47-62)	median 14 (IQR 9-19)	41/57=72% on full or partial sick leave: 20/57=35% partial and 21/57=37% on full sick leave or disability	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Sharif, 2011[48]	1998-?	United States	Longitudinal	patient questionnaire	clinic-based	none	255	212 (83%)	non-WD at baseline: 45.3±13.0, WD at baseline: 50.9±12.5	non-WD at baseline: 2.41±1.61, WD at baseline: 2.71±1.57	124/255=49% working-age were WD at baseline; 35/131=27% became disabled, after mean 4.4±3.8 years of follow-up	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Wilson, 1997[41]	n/a? (~1974-?)	United States	cross-sectional	self-report questionnaire	clinic-based	none	183	86%	n/a?	n/a?	18.8% on disability or sick leave	mean 2.1 days of paid work lost each year	-	mean 11.8 days of unpaid work (among unemployed) lost per year	mean total indirect costs of $10,228 ($8,392 for morbidity and $1,835 for mortality); males: $10,149 overall ($8,393 for morbidity and $1,756 for mortality); females: $10,254 overall ($8,392 for morbidity and $1,862 for mortality) [1994 USD]	mean total indirect costs of $16,815 ($13,796 for morbidity and $3,017 for mortality); males: $16,685 overall ($13,798 for morbidity and $2,887 for mortality); females: $16,858 overall ($13,796 for morbidity and $3,061 for mortality)
Systemic Vasculitis (SV)																
Boomsma, 1999, 2002[51,53][a]	n/a? (~1998)	Netherlands	cross-sectional	self-report questionnaire	clinic-based	clinic-based sample of patients with SLE	79 GPA	35 (44%)	60 (range 27 to 90)	median 5 years (range 0 to 25)	27% receiving disability benefits	-	-	-	-	-
Hoffman, 1998[52]	n/a? (~1997)	United States	cross-sectional	self-report questionnaire	clinic-based	none	60 GPA	47%	54 (range 17-84)	median 5 years (range < 1 to 33)	11/35=31% of once-employed receiving disability benefits	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Mau, 2005[44]	1993-2001	Germany	cross-sectional	self-report questionnaire	clinic-based	German population data	385 GPA	189 (49%)	46 (11)	n/a	SER=0.76 (0.74 for males and 0.79 for females); adjusted RR for employment (vs. RA reference group) of 0.83 (disease duration ≤ 5 years) and 0.62 (disease duration > 10 years)	-	-	-	-	-
Reinhold-Keller, 2002[54]	1996	Germany	cross-sectional	self-report questionnaire	clinic-based	none	60 GPA	34 (57%)	median 36 (range 17 to 48)	median 39 months (range 0 to 228)	16/60=27% unemployed due to GPA	median 14 workdays lost over past 12 months (range 0 to 18)	-	-	-	-

[a]Data from one study were reported across two manuscripts

95% CI=95% confidence interval
dcSSc=diffuse systemic sclerosis
DU=digital ulcers
GPA=Granulomatosus with polyangiitis
IQR=interquartile range
lcSSc=limited cutaneous systemic sclerosis
PsA=psoriatic arthritis
pSjS=primary Sjogren's syndrome
RA=rheumatoid arthritis
RR=relative risk
SER=standardised employment ratio
SLE=systemic lupus erythematosus
sSjS=secondary Sjogren's syndrome

Table 2. Characteristics and Lost Productivity Data for Studies Published from 2012-onwards

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Polymyositis/Dermatomyositis (PM/DM)																
Regardt, 2016[56]	2012	Sweden	cross-sectional	self-report questionnaire or in-person interview	clinic-based	none	48: 23 DM and 25 PM	29 (60%)	54 (10)	9 (9)	12/48=25% on full-time sick leave, 21/48=44% working full-time, 15/48=31% working part-time	-	-	-	-	-
Rice, 2016[1]	1998-2014	United States	cross-sectional	healthcare and disability insurance claims	commercial health insurance beneficiaries aged 18-64 years	sample of beneficiaries without a diagnosis of PM or DM	611 PM/DM and 611 non-PM/DM	64% *among whole cohort, while disability data were only available for a subset*	49.4 (10.6) *among whole cohort, while disability data were only available for a subset*	n/a?	-	medically-related absenteeism: 10.7 days per-year for PM/DM vs. 9.5 for non-PM/DM disability leave: 6.8 days for PM/DM vs. 6.0 for non-PM/DM total work loss: 17.5 days per-year for PM/DM vs. 15.5 for non-PM/DM	-	-	-	-
Sjogren's syndrome (SjS)																
Mandl, 2017[20]	2001-2012	Sweden	Longitudinal	sick leave and disability pensions paid by the Swedish Social Insurance Agency	clinic-based	matched sample of the Swedish general population	51 pSjS and 204 non-SjS	50 (98%)	45.6 (11.3)	n/a (incident cohort)	26% WD at SjD diagnosis; RR (vs. general population) =1.30 (95% CI: 0.74-2.28) 37% WD at 12 months	at diagnosis: 6.2 days of sick leave or disability pension per-month; at 12 months: 9.2 days per-month;	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
											after diagnosis; RR=1.47 (0.83-2.61) 41% WD at 24 months after diagnosis; RR=2.10 (1.34-3.30)	at 24 months: 10.2 days per-month				
Westhoff, 2012[60]	2009-?	Germany	cross-sectional	questionnaire	clinic-based	friends of the SjS cohort, of same sex and similar age	128 pSjS and 84 non-SjS	128 (100%)	50.2 (9.4)	n/a?	10.4% of SjS and 1.5% of non-SjS employed but on sick leave ($p < 0.01$); 28.3% of SjS and 10.7% of non-SjS retired early ($p < 0.01$)	19.8±30.6 days for SjS and 4.5±9 for non-SjS over past 6 months	-	-	-	-
Systemic Sclerosis (SSc)																
Decuman, 2012[58]	2008-2009	Belgium	cross-sectional	self-report questionnaire	clinic-based	none	84	64 (76%)	47.8 (8.9)	56.5 months	47/84=56% made a work transition due to health: 34/84=40% stopped working, 13/84=15% reduced hours and/or changed jobs	-	-	-	-	-
Kawalec, 2015[59]	2012	Poland	cross-sectional	sick leave and disability pensions paid by the Social Insurance Institution of Poland	population-based sample, from among virtually all working patients in the country	population-based samples of SLE and sarcoidosis	500	n/a?	n/a?	n/a?	-	-	-	-	€3341 (Gross Income per worker); €4537 (Gross Domestic Product); €10,927 (Gross Value Added)	$3,876 (Gross Income per worker); $5,052 (Gross Domestic Product);

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
															[2012 Euros]	$12,677 (Gross Value Added)
López-Bastida, 2016[57][a]	2011-2013	France, Germany, Hungary, Italy, Spain, Sweden, United Kingdom	cross-sectional	self-report questionnaire	national and regional patient associations, and Spanish rare diseases registry	none	589 SSc and 57 carers	n/a?	50 (range 45-54)	n/a?	-	-	-	-	<u>Caregivers' time:</u> France: €1,875, Germany: €594, Hungary: €1,050, Italy: €2,812, Spain: €4,684, Sweden: €0, UK: €3,762 <u>Sick Leave:</u> France: €1,501, Germany: €1,369, Hungary: €20, Italy: €790, Spain: €1,445, Sweden: €2,558, UK: €4,912 <u>Early Retirement:</u> France: €8,123, Germany: €10,532, Hungary: €2,241, Italy: €911, Spain: €6,033, Sweden: €0, UK: €9,532 [2012 Euros]	<u>Caregivers' time:</u> France: $2,624, Germany: $831, Hungary: $1,469, Italy: $3,935, Spain: $6,554, Sweden: $0, UK: $5,264 <u>Sick Leave:</u> France: $2,100, Germany: $1,916, Hungary: $1,469, Italy: $1,105, Spain: $2,022, Sweden: $3,579, UK: $6,873 <u>Early Retirement:</u> France: $11,366, Germany: $14,736, Hungary: $3,136, Italy: $1,275, Spain: $8,441, Sweden: $0, UK: $13,337

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Morrisroe, 2016[62]	2007-2015	Australia	cross-sectional	self-report questionnaire	clinic-based	Australian population data	802	670 (84%)	employed: 50.4±10.7; not employed: 51.9±10.4	employed: 9.6±9.0; not employed: 11.1±10.9	160/802=20% not employed	-	-	-	-	-
Sandqvist, 2015[55]	2003-2009	Sweden	longitudinal	sick leave and disability pensions paid by the Swedish Social Insurance Agency	clinic-based	matched sample of the Swedish general population	32 SSc and 128 non-SSc	26 (81%)	median 48 (IQR=43-53)	n/a (incident cohort)	7/32=22% full-time WD after three years (4/8=50% of dcSSc and 3/24=13% of lcSSc); compared to reference group, OR for WD was 0.95 (95% CI: 0.39-2.33) at baseline, 2.09 (1.17-3.73) after one year and 2.41 (1.28-4.55) after three years	mean of 103±130 full or partial days for lcSSc over the first three years, and 190±151 for dSSc	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Singh, 2012[63]	2010?	United States	cross-sectional	self-report questionnaire	clinic-based	none	162	131 (81%)	51.8 (14.2)	7.6 (8.2)	39/162=24% WD due to SSc; 10/43=23% of lcSSc and 24/46=52% of dcSSc	mean 2.6±6.3 days per month among all employed (n=60), 3.5±7.8 for lcSSc and 1.5±2.9 for dcSSc; 24/60=40% missed ≥ 1 work day per month;	mean 2.5±6.1 days of ≤ 50% productivity among all employed (n=60), 3.4±7.5 for lcSSc and 1.2±2.6 for dcSSc; mean 2.2±2.9 days where SSc interfered with productivity among all employed, 2.1±3.0 for lcSSc and 2.7±2.9 for dcSSc;	8.0±10.6 household work days missed among all, 6.2±9.4 for lcSSc and 10.9±11.8 for dcSSc, 8.9 for employed and 6.4 for not employed; 6.0±9.7 days of ≤ 50% productivity for household work among all, 5.5±9.5 for lcSSc and 7.3±10.7 for dcSSc; 2.0±5.0 days with hired help, 1.4±4.0 for lcSSc and 2.7±6.2 for dcSSc 4.0±3.4 days where SSc interfered with household work productivity among all, 3.4±3.3 for lcSSc and 5.1±3.5 for dcSSc	mean income loss of $897 (range $127 to $2,792) per month ($10,764 annually) from absenteeism; $3,577 ±$1,303 per month ($42,924 annually) for WD [2010 USD]	mean income loss of $1,002 per month ($12,024 annually) from absenteeism; $3,996 per month ($47,952 annually) for WD
Systemic Vasculitis (SV)																
Basu, 2014[61]	n/a?	United Kingdom	cross-sectional	self-report questionnaire	clinic-based	none	208: 144 GPA, 40 MPA, 22 EGPA	109 (52.4%)	51.1 (12.3)	n/a?	54/208=26% not working due to health	-	-	-	-	-

Study Characteristics							Characteristics of Study Population				Productivity Outcomes					
First Author, Year	**Study Year(s)**	**Country**	**Design**	**Data Source**	**Study Population / Recruitment Source**	**Comparison Group**	**N Participants**	**N (%) Female**	**Mean (SD) Age, years**	**Mean (SD) Disease Duration, years**	**Work Disability (WD)**	**Paid Work Losses: Absenteeism**	**Paid Work Loses: Presenteeism**	**Unpaid Work Losses**	**Costs (as reported)**	**Costs (2017 USD)**
Barra, 2016[64]	2012-2014	Canada	cross-sectional	self-report questionnaire	clinic-based	none	103: 32 GPA, 12 EGPA, 5 MPA, 24 GCA, 2 Takayasu's, 7 IgA vasculitis, 6 PAN, 4 Behcet's disease, 3 cryoglobulinaemic vasculitis, 2 hypocomplementemic urticarial vasculitis, 1 secondary vasculitis, 5 unclassifiable	60%	58 (17)	4 (4)	22/103=21% (22/51=43% of working age) were WD due to SV: not working (n=13), early retirement (n=3), or working reduced hours (n=6)	-	mean work productivity loss due to health (measured by WLQ): 8.2%	-	-	-

[a]This article reported on costs from each of the seven European countries that participated in this study. The findings from Spain[73] and France[65] are also reported separately.

95% CI: 95% confidence interval
EGPA= Eosinophilic Granulomatosis with polyangiitis (Churg-Strauss syndrome)
GCA=giant cell arteritis
GPA=Granulomatosus with polyangiitis
IQR=interquartile range
MPA=Microscopic polyangiitis
pSjS=primary Sjogren's syndrome
PAN=polyarteritis nodosa
RR=relative risk
WLQ=Work Limitations Questionnaire