

# Economic Burden Associated With Sjögren's Disease (SjD): A Systematic Literature Review (SLR)

Thomas Grader-Beck,<sup>1</sup> Dana DiRenzo,<sup>2</sup> Anjana Lalla,<sup>3</sup> Shweta Takyar,<sup>4</sup> Vinay Pandey,<sup>4</sup> Cindy Chan,<sup>3</sup> Antton Egana<sup>3</sup>

<sup>1</sup>Johns Hopkins School of Medicine, Division of Rheumatology, Baltimore, MD, USA;  
<sup>2</sup>University of Pennsylvania, Division of Rheumatology, Philadelphia, PA, USA;  
<sup>3</sup>Novartis Pharmaceuticals Corporation, East Hanover, NJ, USA;  
<sup>4</sup>Novartis Healthcare Private Ltd., Hyderabad, India

## KEY FINDINGS AND CONCLUSIONS

- This SLR demonstrates that SjD imposes a substantial and growing global economic burden compared with healthy controls, with direct costs 1.5- to 6-fold higher and indirect costs 2- to 3-fold higher. These elevated costs are driven by increased HCRU, medication use, work disability, productivity losses, and higher out-of-pocket expenses
- Greater economic burden was strongly associated with increasing disease severity, systemic organ involvement, and the development of associated conditions, highlighting the importance of early screening, treatment, and multidisciplinary care to mitigate long-term economic strain
- These findings underscore the urgent need for more effective therapeutic strategies that move beyond supportive or symptomatic care and instead focus on disease-modifying treatments

## BACKGROUND

- SjD is a chronic, systemic, heterogeneous, autoimmune disease that can affect multiple organs and is characterized by ocular and oral dryness, debilitating fatigue, and pain<sup>1,2</sup>
- Patients with SjD report high direct-cost burden from frequent outpatient visits, diagnostic testing, and increased medication and dental costs<sup>3,4</sup>
- Additionally, indirect costs can amplify the economic burden of SjD, including lost productivity, lost income from absenteeism, and early retirement, highlighting the substantial socioeconomic impact of SjD<sup>5,6</sup>

## OBJECTIVE

- To identify and summarize published evidence on the global economic burden of SjD, including direct/indirect costs and healthcare resource utilization (HCRU), including comparisons with non-SjD populations

## METHODS

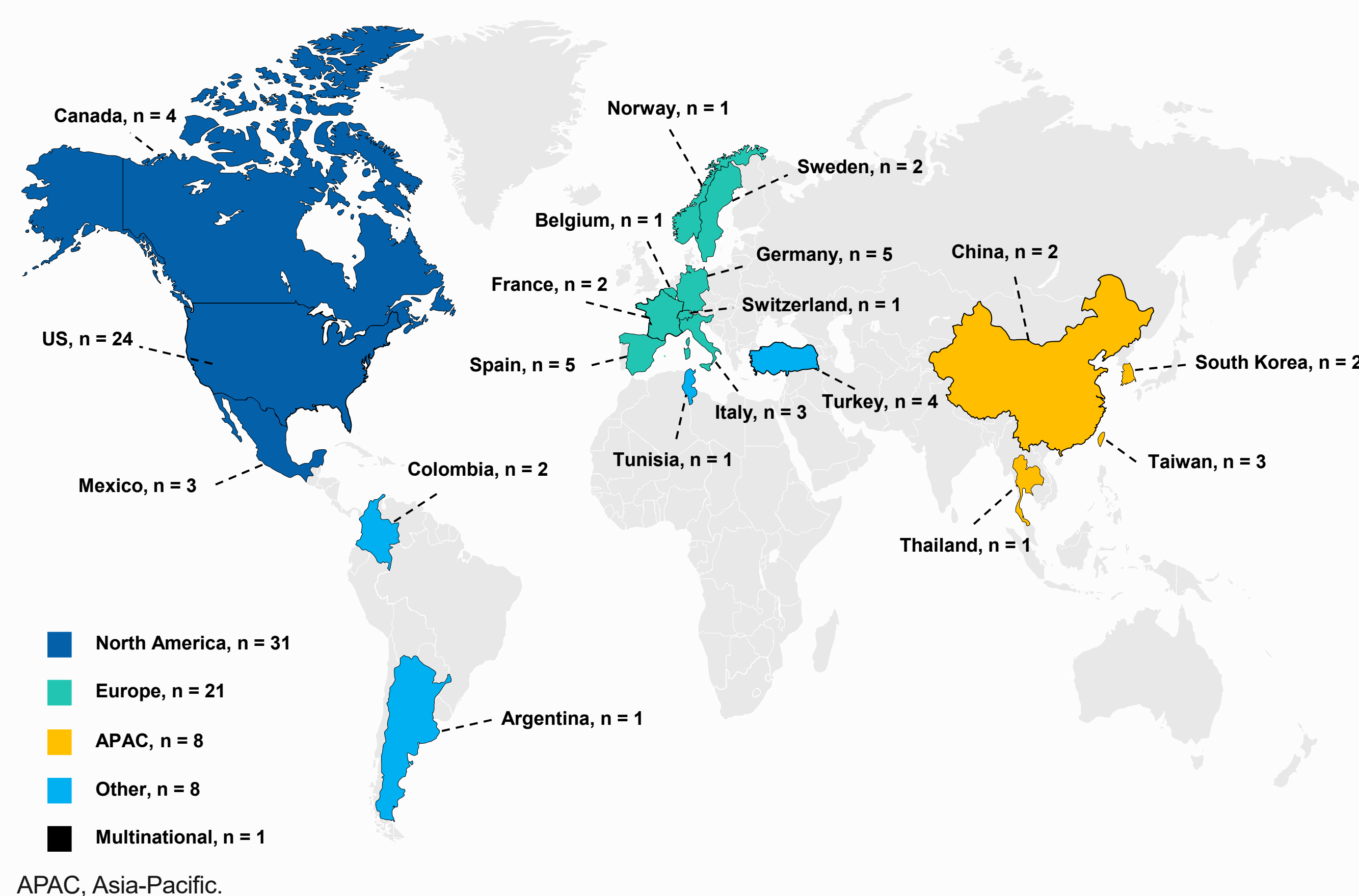
- This SLR was conducted according to established guidelines including National Institute for Health and Care Excellence (NICE) and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Figure 1)
- Key biomedical databases (Embase® and MEDLINE®) were searched between January 2012 and January 2025, and conference proceedings were manually screened from the last 3 years (2022 to 2024)
- The titles and abstracts were screened using pre-defined PICOS inclusion and exclusion criteria (Supplemental Table 1)

## RESULTS

### Study and patient characteristics

- Overall, 70 studies were summarized across all geographies (Figure 2)
  - Mean (SD) age of patients with SjD ranged from 49.3 to 65.1 years,<sup>4,7</sup> women comprised 80% to 100% of patients,<sup>8,9</sup> and most patients were White (62.3%-99.4%)<sup>10,11</sup>
  - The most frequently reported SjD symptoms were ocular and oral dryness (93%); glandular symptoms and fatigue were also common<sup>4</sup>
  - Treatment strategies varied with high use of symptomatic treatments, such as artificial tears (97%),<sup>4</sup> and systemic therapies including corticosteroids (91%)<sup>12</sup> and hydroxychloroquine (54.5%-87.4%)<sup>11,13</sup>

### Figure 2. Number of Included Studies by Geographic Distribution



### Direct costs<sup>3-6,14-17</sup>

- 24 studies reported direct costs, of which 6 included comparisons with non-SjD controls (Table 1); the average total direct costs were 1.5 to 6x higher among patients with SjD than non-SjD
  - Direct costs ranged from \$5748 to \$27,526 in the US; higher total annual costs were reported in Europe in SjD vs non-SjD populations (≈3x higher in Italy; ≈1.5x higher in Sweden)
  - Key direct cost drivers were medical visits in the US (≈80% of all-cause costs), including hospitalizations and outpatient services, pharmacotherapy in Europe (3x higher among patients with SjD vs controls), and out-of-pocket (OOP) spending due to prescription medications and dental care in Sweden (1.4x higher than controls)
  - Higher costs were linked with associated SjD (SjD + autoimmune diseases; 1.4 to 1.5x higher) vs primary SjD (SjD alone), extraglandular disease (2.9x higher) vs glandular disease, interstitial lung disease (ILD; 2.7x higher) vs no ILD, and SjD with acute renal injury (11% higher) vs acute renal injury alone

## References

- Singh AG, et al. *Rheumatology (Oxford)*. 2016;55(3):450-460.
- Lee AYS, et al. *Arthritis Care Res (Hoboken)*. 2023;75(7):1580-1587.
- Perera S, et al. *J Health Econ Outcomes Res*. 2018;5(2):150-161.
- McCoy SS, et al. *Clin Rheumatol*. 2022;41(7):2071-2078.
- Westerlund A, et al. *Rheumatol Ther*. 2021;8(2):955-971.
- Chan C, et al. *BMJ Open Ophthalmol*. 2021;6(1):e000709.
- Falasinu T, et al. *Lancet Rheumatol*. 2024;6(8):e518-e527.
- Foocharoen C, et al. *Int J Rheum Dis*. 2013;16(1):41-46.
- Dias LH, et al. *Ann Rheum Dis*. 2019;74:812-812.
- Falasinu T, et al. *ACR Open Rheumatol*. 2022;4(10):890-896.
- Meinecke A, et al. *Clin Exp Rheumatol*. 2024;42(12):2444-2452.
- Callhoff J, et al. *Clin Exp Rheumatol*. 2019;37:S83-S89.
- Yayla ME, et al. *Clin Rheumatol*. 2022;41(5):1457-1463.
- Kalouche-Khalil L, et al. *Annals of the Rheumatic Diseases*. 2022;81:410-411.
- McCormick N, et al. *J Rheumatol*. 2015;42(7):13-25.
- Cafaro G, et al. *Eur J Intern Med*. 2024;124:122-129.
- Yao W, et al. *BMC Ophthalmol*. 2018;18(1):23.
- McCormick N, et al. *Arthritis Care Res*. 2019;71(1):142.
- McCoy S, et al. *Arthritis Rheumatol*. 2023;75 (suppl 9).
- Ndlfe B, et al. *Arthritis Rheumatol*. 2021; 73 (suppl 9).

## Figure 1. PRISMA Flow Diagram of Included Studies

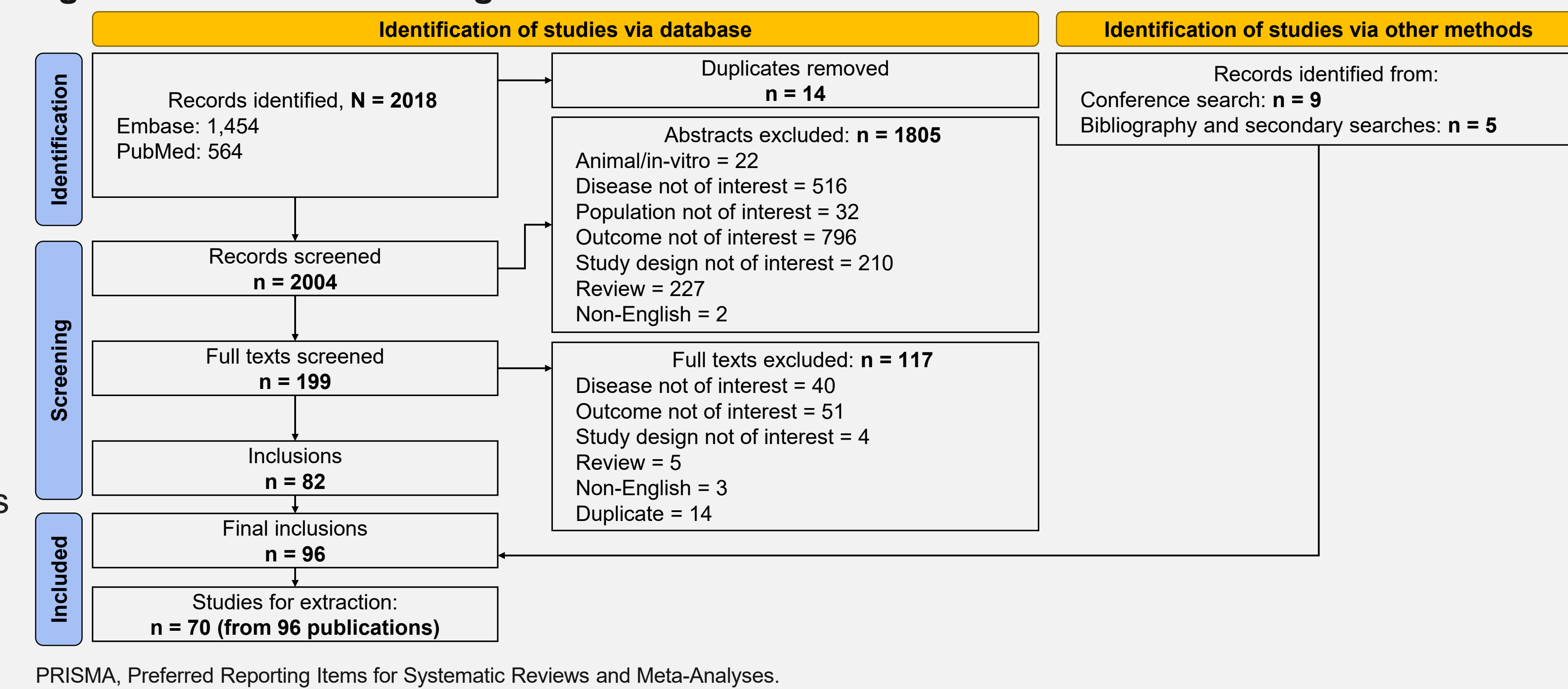


Table 1. Total Direct Costs in Patients With SjD vs the General Population or Non-SjD Controls

Study name	Country	SjD type	Study population	Cost year	Cost type	Unit	Costs	P value
Perera 2018 <sup>3</sup>	US	Primary	SjD	2013	Total all-cause cost	PPPY, mean	USD 10,729	-
		NA	non-SjD (sicca-free)				USD 6534	
		Primary	SjD				USD 2214	
McCormick 2015 <sup>15</sup>	Canada	Overall	SjD	2010	Total cost	PPPY, mean (IQR)	CAD 11,748 (9640-13,856)	<.01
		NA	General population				CAD 4213 (3762-4,665)	
		Overall	SjD (with DED)				CAD 2689 (2,430)	
Chan 2021 <sup>6</sup>	Sweden	NA	non-SjD (with DED)	2018	Total cost	PPPY, mean (SD)	CAD 2203 (6,660)	-
		Primary	SjD				SEK 51,384 (80,423)	
Westerlund 2021 <sup>5</sup>	Sweden	NA	General population	2016	Total cost	Total cost per patient, mean (SD)	SEK 28,497 (64,683)	<.0001
		Primary	SjD				SEK 28,497 (64,683)	
Cafaro 2024 <sup>16</sup>	Italy	Overall	SjD	2018	Total cost	PPPY, mean	€1171	<.01
		NA	non-SjD				€372	
Yao 2018 <sup>17</sup>	China	Overall	SjD (with DED)	2016	Total cost	PPPY, mean (SD)	CNY 7637 (6079)	<.001
		NA	non-SjD (with DED)				CNY 1179 (990)	

CAD, Canadian dollars; CNY, Chinese yuan renminbi; DED, dry eye disease; IQR, interquartile range; NA, not applicable; PPPY, per patient per year; SD, standard deviation; SEK, Swedish krona; SjD, Sjögren's disease; USD, US dollars.

### Indirect costs<sup>6,17,18</sup>

- 3 studies reported indirect costs compared with non-SjD controls (Table 2); indirect costs were 2 to 3x higher for patients with SjD
  - Key indirect cost drivers like presenteeism were more dominant than absenteeism, accounting for 64% of total paid work costs in SjD vs 53% in non-systemic autoimmune rheumatic diseases (SARDs); patients with SjD also had higher unpaid work losses than patients without SARDs (60% vs 47% of total productivity costs)
  - Risk factors for increased indirect costs included overweight status, functional disability, pain, and fatigue

Table 2. Total Indirect Costs in Patients With SjD vs the General Population or Non-SjD Controls

Study name	Country	SjD type	Study population	Cost year	Cost type	Unit	Costs	P value
Chan 2021 <sup>6</sup>	Canada	Overall	SjD (with DED)	2018	Productivity loss	PPPY, mean	CAD 41,094	NR
		NA	non-SjD (with DED)				CAD 17,694	
McCormick 2019 <sup>18</sup>	Canada	Overall	SjD	2015	Productivity loss	PPPY	CAD 19,599	NR
		NA	non-SARD				CAD 11,144	
Yao 2018 <sup>17</sup>	China	Overall	SjD (with DED)	2016	Medication expense <sup>a</sup>	PPPY, mean	CNY 828	.017
		NA	non-SjD (with DED)				CNY 487	

CAD, Canadian dollar; CNY, Chinese yuan renminbi; NA, not applicable; NR, not reported; SARD, systemic autoimmune rheumatic disease; SjD, Sjögren's disease; PPPY, per patient per year.

<sup>a</sup>Indirect costs for activities associated with medical treatment, such as transportation, nutrition, shelter, and household appliance.

### Healthcare resource utilization

- 49 studies reported HCRU; patients with SjD experienced 1.15 to 3x higher HCRU vs non-SjD controls (Supplemental Table 2) characterized by increased hospitalization rates, frequent outpatient visits, increased multidisciplinary care, and greater pharmacological use

### Work productivity and activity impairment (WPAI)<sup>19,20</sup>

- 25 studies reported WPAI and found consistently higher average rates of absenteeism, presenteeism, and activity impairment than controls (Supplemental Table 3)
  - Presenteeism consistently outweighed absenteeism (29.8% vs 5.0%)
  - Disease severity was strongly associated with higher WPAI in the US (Figure 3); diagnostic delays ≥12 months led to greater work impairment

### OOP costs<sup>4</sup>

- A US-based survey reported 35% of annual OOP costs (\$2026) were for dental care, followed by prescription medications (\$1319) and over-the-counter/alternative therapies (\$1290), underscoring a substantial patient burden

Figure 3. Physician-Perceived Disease Severity and Associated WPAI Among Patients With SjD in the US<sup>20</sup>

