

Supplemental Material

Supplemental Table 1. Inclusion and exclusion criteria of this SLR

Criteria	Inclusion criteria	Exclusion criteria
Population	Adult patients with active SjD Any study retrieved in the search timeframe reporting for adult primary/secondary Sjogren's patients will also be included	All other diseases Patients aged <18 years (children and adolescents)
Interventions/comparators	No restriction on intervention and comparator	N/A
Outcomes	<p>Economic burden</p> <ul style="list-style-type: none"> • Direct costs (e.g. drug costs, outpatient costs, inpatient costs, emergency department visit costs etc.) • Indirect costs (cost associated with productivity loss due to presenteeism or absenteeism etc.) • Healthcare resource utilization (number of outpatient visits, inpatient visits, emergency department visits, length of stay etc.) • Work-productivity loss (productivity impairment, activity impairment, decreased number of hours etc.) • Caregiver burden (caregiver strain etc.) 	Studies not reporting data pertaining to economic burden of SjD
Study designs	Observational studies (prospective and retrospective cohort, case-control studies, cross-sectional studies, longitudinal studies etc.)	Prospective interventional studies
Publication timeframe	2012 onwards	N/A
Language	Articles in English	Non-English studies
Country	No country restriction	N/A

N/A, not applicable; SjD, Sjögren's disease; SLR, systematic literature review.

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Supplemental Table 2. HCRU in patients with SjD vs the general population or non-SjD controls

Study name	Country	SjD type	Population	Sample size	HCRU type	HCRU period	No. of patients with HCRU (n)	Proportion of patients with HCRU (%)	Unit	Mean (SD)	HCRU %	P value			
Maciel 2018 ¹	USA	Primary	SjD	160	Hospitalization	1995-2016	-	-	Rate per 100 person-years	24.2	-	-			
		NA	General population	466			-	-		19.3	-	-			
		Primary	SjD	160	Length of stay		-	-	Per patient	3.9	-	-			
		NA	General population	466			-	-		4.7	-	-			
		Primary	SjD	160	Readmissions		-	18% of subsequent hospitalizations	Per cohort	-	-	-			
		NA	General population	466			-	19% of subsequent hospitalizations		-	-	-			
Perera 2018 ²	USA	Primary	SjD	12,717	SjD related - outpatient hospital	2013	-	-	Per patient	2.9	-	-			
		NA	Non-SjD (sicca-free)							2.6	-	-			
		Primary	SjD		SjD related - inpatient hospital					1.2	-	-			
		NA	Non-SjD (sicca-free)							1.1	-	-			
		Primary	SjD		SjD related - hospital emergency room					1.2	-	-			
		NA	Non-SjD (sicca-free)							1.2	-	-			
		Primary	SjD		SjD related - urgent care facility					1.2	-	-			
NA	Non-SjD (sicca-free)	1.1	-	-											
McCormick 2015 ³	Canada	Overall	SjD	691	Hospitalization in first year after diagnosis	1996-2008	200	29	-	-	-				
Cafaro 2024 ⁴	Italy	Primary	SjD	1746	Hospitalizations: ≥ 1	2018	137	7.8	Per patient	1.5	-	<.001			
		NA	Controls (without SjD)				68	3.9		1.3	-	-			
		Primary	SjD		Length of in-hospital stay (days)		-	-		14.3	-	<.001			
		NA	Controls (without SjD)				8.4	-		-					
		Primary	SjD		Outpatient specialist services: ≥ 1		869	49.8		42.7	-	-			
		NA	Controls (without SjD)				534	30.6		21.9	-	-			
		Primary	SjD		Pharmacological treatments: ≥ 1		937	53.7		1282.3	-	-			
NA	Controls (without SjD)	746	42.7	685.8		-	-								
Albrecht 2020 ⁵	Germany	Primary	SjD	2509	Total hospitalizations	2018	803	32	-	-	-	-			
		Associated	SjD	3457			1348	39		-	-				
		NA	Controls (without SjD)	29,805			6259	21		-	-				
		Both	Primary SjD	2509			2007	853		34	-	-			
		Both	Associated SjD	3457				1175		34	-	-			
		NA	Controls (without SjD)	29,805				4769		16	-	-			
Westhoff 2012 ⁶	Germany	Primary	SjD	176	Physician visits	6 months	-	-	Per patient	17 (10)	-	<.001			
		NA	Controls (without SjD)	115						6.5 (4.5)	-	-			
		Primary	SjD	176	Specialist visits ^a				Per patient (range)	0.4-4.2	-	-			
		NA	Controls (without SjD)	115						0.1-1.6	-	-			
		Primary	SjD: ≤ 64 years	128	Psychotherapy				2009	-	-	-	35.8	-	-
		NA	Controls (without SjD): ≤ 64 years	84									15.8	-	-
		Primary	SjD: ≤ 64 years	128	Currently in psychotherapeutic treatment				-	-	-	-	12.1	-	-
		NA	Controls (without SjD): ≤ 64 years	84									2.6	-	-
		Primary	SjD: ≤ 64 years	128	Antidepressants				-	-	-	-	16.1	-	-
		NA	Controls (without SjD): ≤ 64 years	84									6.2	-	-

HF, heart failure; HCRU, healthcare resource utilization; NA, not applicable; SjD, Sjögren's disease.

^a Specialist visits include: rheumatologist, orthopedic surgeon, ophthalmologist, ENT specialist, dermatologist, neurologist, family practitioner, surgeon, gynecologist, dentist and others.

1. Maciel G, et al. *RMD Open*. 2018;4(1):e000575. 2. Perera S, et al. *J Health Econ Outcomes Res*. 2018;5(2):150-161. 3. McCormick N, et al. *J Rheumatol*. 2015;42(7):1325. 4. Cafaro G, et al. *Eur J Intern Med*. 2024;124:122. 5. Albrecht K, et al. *Clin Exp Rheumatol*. 2020;38 Suppl 126(4):78-84. 6. Westhoff G, et al. *Rheumatology (Oxford)*. 2012;51(2):262-269.

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Supplemental Table 2 (cont). HCRU in patients with SjD vs the general population or non-SjD controls

Study name	Country	SjD type	Population	Sample size	HCRU type	HCRU period	No. of patients with HCRU (n)	Proportion of patients with HCRU (%)	Unit	Mean (SD)	HCRU %	P value
Westerlund 2021⁷	Sweden	Primary	SjD	7396	Total hospitalizations	2015	-	-	Per patient	0.5 (1.2)	-	<.0001
			NA	General population						76,624		0.3 (0.9)
		Primary	SjD	7396	Length of in-hospital stay (days)					2.6 (9.4)		<.0001
			NA	General population						76,624		0
		Primary	SjD	7396	Outpatient visits					4.4 (6.3)		<.0001
			NA	General population						76,624		2 (4.1)
		Primary	SjD	7396	Dental visits					2.3 (2.3)		<.0001
			NA	General population						76,624		1.6 (1.9)
		Primary	Overall SjD	7396	Dental procedures					3.8 (4)		<.0001
NA	General population		76,624	2.8 (3.3)		-						
Chen 2015⁸	Taiwan	Overall	SjD	4287	Hospitalizations	1996-2011	-	5.6	-	-	-	
			Control	25,722				2.6				
Lin 2022⁹	Taiwan	Primary	Overall SjD	16,466	Incident hospitalization events due to HF at 1 year	2000-2013	-	Per cohort	-	0.3	-	
										Incident hospitalization events due to HF at 3 years	1.05	
										Incident hospitalization events due to HF at 5 years	1.89	
										Incident hospitalization events due to HF at 10 years	4.33	
										Incident hospitalization events due to HF	0.27	
										Incident hospitalization events due to HF at 1 year	1.1	
										Incident hospitalization events due to HF at 3 years	1.89	
										Incident hospitalization events due to HF at 5 years	4.44	
										NA	Controls (without SjD)	

HF, heart failure; HCRU, healthcare resource utilization; NA, not applicable; SjD, Sjögren's disease.

^a Specialist visits include: rheumatologist, orthopedic surgeon, ophthalmologist, ENT specialist, dermatologist, neurologist, family practitioner, surgeon, gynecologist, dentist and others.

7. Westerlund A, et al. *Rheumatol Ther.* 2021;8(2):955-971. 8. Chen JY, et al. *PLoS One.* 2015;10(8). 9. Lin CY, et al. *Ther Adv Chronic Dis.* 2022;13(1).

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Supplemental Table 3. Work productivity and activity impairment in patients with SjD versus the general population or non-SjD controls

Study name	Country	SjD type	Population	Sample size	HCRU item	Resource use period	Number of patients with HCRU (n)	Proportion of patients with HCRU (%)	Unit	Mean (SD) HCRU	P value
McCormick 2019 ¹	Canada	Overall	SjD: employed	33	Absenteeism: hours worked, past 7 days, paid work	1996 - 2010	12	36	Per cohort	4 (8.2)	.62
		NA	Non-SARD: employed	160			26	16		3.1 (9.6)	-
		Overall	SjD: employed	33	Presenteeism: hours worked, past 7 days, paid work		25	83		7.6 (8.6)	.01
		NA	Non-SARD: employed	160			77	52		4.1 (6.2)	-
		Overall	SjD: employed	33	Absenteeism and presenteeism, hours worked, 7 days, paid work		27	82		10.9 (11.6)	.08
		NA	Non-SARD: employed	160			84	53		6.9 (11.8)	-
		Overall	SjD	90	Productivity loss, hours, past 7 days, unpaid work		40	44		6.4 (13.1)	<.01
		NA	Non-SARD	375			93	25		2.6 (7.5)	-
		Overall	SjD	90	Productivity loss, hours, past 7 days, paid and unpaid work		53	59		10.4 (15.9)	<.01
		NA	Non-SARD	375			148	39		5.5 (12)	-
Albrecht 2020 ²	Germany	Primary	SjD: ≤ 65 years	1173	Persons with sick leave	2018	562	48	Per patient	-	-
		Associated	SjD: ≤ 65 years	1474			642	44		-	-
		NA	Matched controls: ≤ 65 years	13,215			5419	41		-	-
		Primary	SjD: ≤ 65 years	1173	-		-	53		-	
		Associated	SjD: ≤ 65 years	1474	Sick leave, days		-	-		67	-
		NA	Matched controls: ≤ 65 years	13,215			-	-		40	-

HCRU, healthcare resource utilization; NA, not applicable; NR, not reported; SARD, systemic autoimmune rheumatic diseases; SjD, Sjögren's disease.

1. McCormick N, et al. *Arthritis Care Res (Hoboken)*. 2019;71(1):142. 2. Albrecht K, et al. *Clin Exp Rheumatol*. 2020;38 Suppl 126(4):78-84.

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Supplemental Table 3 (cont). Work productivity and activity impairment in patients with SjD versus the general population or non-SjD controls

Study name	Country	SjD type	Population	Sample size	HCRU item	Resource use period	Number of patients with HCRU (n)	Proportion of patients with HCRU (%)	Unit	Mean (SD) HCRU	P value
Westhoff 2012 ³	Germany	Primary	SjD: ≤ 64 years	67	Working hours per week	2009	-	-	Per patient	35.2 (9.3)	.737
		NA	Controls: ≤ 64 years	64			-	-		34.6 (10.6)	-
		Primary	SjD: ≤ 64 years	67	On sick leave at present		7	10.4		-	<.001
		NA	Controls: ≤ 64 years	84			1	1.5		-	-
		Primary	SjD: ≤ 64 years	67	Days of sick leave in the past 6 months		-	-		19.8 (30.6)	<.001
		NA	Controls: ≤ 64 years	84			-	-		4.5 (9)	-
		Primary	SjD: ≤ 64 years	128	Early retired		36	28.3		-	<.001
		NA	Controls: ≤ 64 years	84			9	10.7		-	-
		Primary	SjD: ≤ 64 years	67	Self-rated working prospects poor		30	44.8		-	<.001
		NA	Controls: ≤ 64 years	64			3	4.7		-	-
		Primary	SjD: ≤ 64 years	128	Unemployed at present		10	7.9		-	.001
		NA	Controls: ≤ 64 years	84			2	2.4		-	-
		Primary	SjD: ≤ 64 years	128	Not working at present		50	39.1		-	<.001
		NA	Controls: ≤ 64 years	84			17	17.9		-	-
Westerlund 2021 ⁴	Sweden	Primary	SjD	2804		5 years after index date (prevalent analysis)	745	26.5		-	-
		NA	General population	34,325			3,968	11.5		-	-
		Primary	SjD	NR	Early retirement	At index date	-	23.4		-	-
		NA	General population	NR			-	12.1		-	-
		Primary	SjD	NR		10-year post index date (incident analysis)	-	51.6		-	-
		NA	General population	NR			-	1.8		-	-

HCRU, healthcare resource utilization; NA, not applicable; NR, not reported; SARD, systemic autoimmune rheumatic diseases; SjD, Sjögren's disease.

3. Westhoff G, et al. *Rheumatology (Oxford)*. 2012;51(2):262-269. 4. Westerlund A, et al. *Rheumatol Ther*. 2021;8(2):955-971.