# Economic Burden of Interstitial Lung Disease in a Commercially Insured Population with Sjögren's Syndrome in the United States

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## Background and Objective

- Patients with Sjogren's syndrome (SjS) have substantial cost burden on the healthcare system.
- Among these patients, those who develop interstitial lung disease (ILD) suffer from poorer quality of life and risk higher mortality.
- The economic burden of interstitial lung disease has not yet been documented.
- Our aim was to estimate the direct healthcare costs associated with ILD among patients with SjS in a representative sample of the commercially insured population in the United States.

## Methods

- Sample population and data source: Commercially insured beneficiaries diagnosed with SjS between January 1, 2006, and September 30, 2015, with or without a diagnosis of ILD, were identified using the PharMetrics Plus for Academics database.
- Selection criteria:
  - 180-day pre- and post-index continuous enrollment
  - Age 18 or older
- Follow up: 180 days post index
- Index date: the later date of the first claim with a diagnosis of SjS or the first claim with a diagnosis of ILD for SjS-ILD cases, and the first claim with a diagnosis of SjS for SjS-only controls.
- Statistical analysis:
  - 5:1 propensity score (PS) matching controlling for baseline demographic and geographic variables
  - Descriptive statistics and generalized linear model of total cost with covariate-adjusted cost ratio and adjusted average marginal cost reported

## Results

- Final sample: 815 SjS-ILD cases and 4,075 PS-matched SjS-only controls.
- Table 1 illustrates the demographic and clinical characteristics of the matched sample.
- The mean (SD) 180-day total costs were $23,192 ($48,189) for SjS-ILD cases and $8,648 ($17,478) for SjS-only controls.
- Table 2 suggests that the main cost drivers were outpatient services other than physician office visit (such as radiological and pathological tests), inpatient services, and outpatient pharmacy cost components for both groups.
- Healthcare costs were doubled among patients with SjS who had ILD compared to those who did not have ILD. The additional six-month cost was $8,814 on average. (Table 2)

**Table 2.** Mean 180-day costs in the SjS-ILD cohort and the SjS-only cohort among a total of 4,890 PS-matched subjects

| | SjS-ILD cases (n=815) | | | PS-matched[a] SjS-only controls (n=4,075) | | | Adjusted Cost Ratio (95% CI)[b,c] | Adjusted Average Marginal Cost (95% CI)[b,c,d] |
|---|---|---|---|---|---|---|---|---|
| | Mean (SD) | % | Median (IQR) | Mean (SD) | % | Median (IQR) | | |
| **Total cost** | $23,192 ($48,189) | 100.0% | $7,960 ($18,668) | $8,648 ($17,478) | 100.0% | $3,369 ($7,107) | 1.95 (1.76-2.15)* | $8,814 ($7,149-$10,479)* |
| **Cost per HCRU category** | | | | | | | | |
| **Inpatient** | $8,997 ($36,824) | 38.8% | $0 ($1,532) | $1,952 ($11,769) | 22.6% | $0 ($0) | n/a | n/a |
| **ED visit** | $232 ($964) | 1.0% | $0 ($0) | $140 ($645) | 1.6% | $0 ($0) | n/a | n/a |
| **Pharmacy** | $3,785 ($8,212) | 16.3% | $1,273 ($3,575) | $1,977 ($4,472) | 22.9% | $540 ($1,879) | n/a | n/a |
| **Physician office visit** | $1,036 ($1,076) | 4.5% | $720 ($1,031) | $799 ($921) | 9.2% | $545 ($752) | n/a | n/a |
| **Other outpatient[e]** | $9,142 ($21,304) | 39.4% | $2,929 ($7,453) | $3,781 ($8,723) | 43.7% | $1,062 ($3,177) | n/a | n/a |

[a]Variables that are used as independent variables in the PS model include the following: age, sex, residence region, index year, and plan product type.
[b]GLM with a gamma distribution and a log link was fit. Costs represented strictly to non-zero cost to fit gamma distribution (N=4,836; 811 cases and 4,025 controls).
[c]Independent variables included in the GLM were comparison group (i.e., SjS-ILD versus SjS-only), age, dummy variables for index year categories, dummy variables for CCI categories, baseline RA, baseline SLE, and baseline systemic sclerosis.
[d]Confidence interval was estimated using the delta method.
[e]Other outpatient services refer to outpatient services other than physician office visits, including laboratory, pathology, radiology, outpatient surgical, and ancillary services.
* p-value <0.01
ED = emergency department; GLM = generalized linear model; HCRU = health care resource utilization; ILD = interstitial lung disease; IQR = interquartile range; PS = propensity-score; RA = rheumatoid arthritis; SD = standard deviation; SjS = Sjogren's syndrome; SLE = systemic lupus erythematosus.

## Conclusions

- Total direct healthcare cost was substantially higher in patients with SjS and ILD compared to patients with SjS without ILD.
- Our findings provide the foundation for further economic evaluation for preventive strategies to reduce the clinical and economic burden imposed by ILD among patients with SjS.

## Results (cont.)

**Table 1.** Demographic and clinical characteristics of commercially insured beneficiaries diagnosed with SjS with and without ILD after PS-matching from 2006 – 2015 (N = 4,890)

| Characteristics | SjS-ILD cases (n=815) | PS-matched SjS-only controls (n=4075) | P value[a] | Standardized difference |
|---|---|---|---|---|
| **Age, yrs, mean (SD)** | 58.47 (11.43) | 53.47 (12.32) | <0.01 | 0.41 |
| **Sex, n (%)** | | | 0.20 | 0.05 |
| **Female** | 696 (85.4) | 3548 (87.07) | | |
| **Male** | 119 (14.6) | 527 (12.93) | | |
| **Residence region, n (%)** | | | 0.90 | 0.03 |
| **Northeast** | 197 (24.17) | 991 (24.32) | | |
| **Midwest** | 212 (26.01) | 1019 (25.01) | | |
| **South** | 288 (35.34) | 1442 (35.39) | | |
| **West** | 118 (14.48) | 623 (15.29) | | |
| **Index year, n (%)** | | | <0.01 | 0.13 |
| **2006-2008** | 160 (19.63) | 1012 (24.83) | | |
| **2009-2011** | 330 (40.49) | 1478 (36.27) | | |
| **2012-2015** | 325 (39.88) | 1585 (38.9) | | |
| **Plan product type, n (%)** | | | 0.44 | 0.06 |
| **Preferred provider organization** | 608 (74.6) | 3050 (74.85) | | |
| **Health maintenance organization** | 102 (12.52) | 467 (11.46) | | |
| **Others/unknown[b]** | 105 (12.88) | 558 (13.69) | | |
| **Quan Charlson comorbidity index, categories** | | | <0.01 | 1.02 |
| **0** | 165 (20.25) | 2550 (62.58) | | |
| **1** | 301 (36.93) | 962 (23.61) | | |
| **2+** | 349 (42.82) | 563 (13.82) | | |
| **Other related autoimmune diseases[c]** | | | | |
| **Rheumatoid arthritis** | 161 (19.75) | 460 (11.29) | <0.01 | 0.24 |
| **Systemic lupus erythematosus** | 125 (15.34) | 264 (6.48) | <0.01 | 0.29 |
| **Systemic sclerosis** | 58 (7.12) | 69 (1.69) | <0.01 | 0.27 |

[a] Student's t-test was used for continuous variables, chi-square test was used for categorical variables, and Cochran-Mantel-Haenszel test was used for multi-level nominal variables.
[b] Other health plan product types include Consumer Directed Health Care, Indemnity Plan, and Point of Service.
[c] Dermatomyositis, and polymyositis were not included due to rare or zero prevalence in our sample.

## Implications

ILD is irreversible and causes clinical and economic burdens. Treatments are available that can slow down the disease progression. To inform formulary decisions, this study is the necessary first step to quantify the cost of illness.

## Contact Information

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