SARCOIDOSIS VASCULITIS AND DIFFUSE LUNG DISEASES 2020; 37 (3); e2020002 DOI: 10.36141/svdld.v37i3.9261

Patterns of healthcare resource utilization in patients with sarcoidosis: a cross-sectional study

Nynke A. Kampstra[1], Paul B. van der Nat[2], Frouke T. van Beek[3], Jan C. Grutters[4], Douwe H. Biesma[5], Philip J. van der Wees[6]

[1] Department of Value-Based Healthcare, St. Antonius Hospital, Santeon-group. Radboud university medical center , Radboud Institute for Health Sciences, Scientific Center for Quality of Healthcare (IQ healthcare), Nijmegen, the Netherlands; [2] Department of Value-Based Healthcare, St. Antonius Hospital, Nieuwegein, the Netherlands. Radboud university medical center, Radboud Institute for Health Sciences, Scientific Center for Quality of Healthcare (IQ healthcare), Nijmegen, the Netherlands; [3] Interstitial Lung Diseases Center of Excellence, Department of Pulmonology, St. Antonius Hospital, Nieuwegein, the Netherlands; [4] Interstitial Lung Diseases Center of Excellence, Department of Pulmonology, St. Antonius Hospital, Nieuwegein, the Netherlands. Division of Heart and Lungs, University Medical Centre Utrecht, Utrecht, The Netherlands; [5] Department of Value-Based Healthcare, St. Antonius Hospital, Santeon-group Department of Internal Medicine, University Medical Centre Utrecht, The Netherlands, St. Antonius Hospital, Nieuwegein, the Netherlands; [6] Radboud university medical center, Radboud Institute for Health Sciences, Scientific Center for Quality of Healthcare (IQ healthcare), Department of Rehabilitation.

Abstract. *Background:* Limited data are available on healthcare resource use and costs in patients with sarcoidosis; *Objectives:* The primary aim of this study was to describe cost-drivers of the top 1% and top ≥1-5% high-cost patients with sarcoidosis. The secondary aim was to compare costs of patients with and without fatigue complaints and to compare comorbidities. *Methods:* We conducted a retrospective observational cross-sectional study in 200 patients diagnosed with sarcoidosis. Hospital administrative databases were used to extract healthcare utilization on the individual patient level. Healthcare costs were categorized into nine groups. *Results:* Average total health care costs for the top 1% (n=22), top ≥1%–5% (n=88) and bottom 95% beneficiaries (n=90) were € 108.296, €53.237 and €4.817, respectively. Mean treatment time in days for the top 1%, top ≥1-5% and the random sample of the bottom 95% was 1688 days (±225), 1412 days (±367) and 775 days (±659), respectively. Mean annual costs for the top 1%, top ≥1-5% and the random sample of the bottom 95% are €51.082, €27.840 and €8.692, respectively. We identified three cost-drivers in the top 5% high-cost patients: 1) expensive medication, 2) intensive care and 3) costs made at the respiratory unit. Patients with and without fatigue showed to have comparable mean costs. High-cost patients were more likely to have multiple organs involved due to sarcoidosis. *Conclusions:* We identified expensive medication as the main cost-driver in the top 5% high-cost patients with sarcoidosis. The study findings can help to tailor interventions for improving the quality of care and reducing overall costs. *(Sarcoidosis Vasc Diffuse Lung Dis 2020; 37 (3): e2020002)*

Key words: sarcoidosis, costs, quality of care

Introduction

Sarcoidosis is a chronic granulomatous disease characterized by persistent body complaints in multiple organs and patients suffer from a broad range of nonspecific symptoms. (1–4) The inflammation level as well as organs affected are highly variable. In more than 90% of the cases, sarcoidosis affects the lungs. Health-related quality of life (HRQoL) and

Received: 7 February 2020
Accepted after revision: 17 August 2020
Correspondence: Nynke A. Kampstra
Koekoekslaan 1, 3435 CM Nieuwegein, The Netherlands
E-mail: n.kampstra@antoniusziekenhuis.nl
or nynkekampstra@gmail.com

health status is often reduced in patients with sarcoidosis. (5–8) Fatigue is an often reported symptom in patients with sarcoidosis. (2, 9, 10) In a sample of 1197 patients with sarcoidosis, 70% of the patients reported fatigue as a feature of their sarcoidosis. (10)

Detailed insights into the costs of patients with advanced sarcoidosis are however lacking. Efforts to further improve the quality of care could specifically target advanced sarcoidosis patients. Therefore, it would be useful to have a detailed picture of high-cost patients with sarcoidosis which can enable better evaluation of the current treatment choices.

The most complex patient group is that with the highest costs and needs, with a variety of complex medical conditions. About 5% of patients with various conditions account for 50% of the total health care spending. (11) Detailed insight into the disease specific costs regarding patients with sarcoidosis are getting more attention in the literature, although this is still limited. (12–14) Previous research has demonstrated that the cost distribution for patients with sarcoidosis is highly skewed. High-cost patients had more sarcoidosis related comorbidities compared to low-cost patients. (12–14) Previous research has shown commercial payers in the USA incurred a mean of $19,714 total annual healthcare costs per sarcoidosis patient. (12) It was furthermore reported that the main cost-drivers identified were outpatient visits (46% of the costs) and inpatient admissions (32% of the costs). Mean health care costs in a US-based patient group in the top quintile were $73,346. The authors furthermore concluded that this subgroup of most costly patients with sarcoidosis might be worthwhile to invest in concerning outcome improvement efforts. Another study, based on a U.S. national health care database, found that the median healthcare costs were $18,663 for patients with sarcoidosis per year. (14) The top 5% costliest patients with sarcoidosis exceeded $90,000 per year. However, little is known on the health care utilization, the patient characteristics and the relationship with fatigue complaints in high-cost patients with sarcoidosis. Furthermore, no other studies have tried to identify cost-drivers in this high-cost patient subgroup and identify their patient characteristics in a (Dutch) cohort. This can be valuable as it will enable to assess the effectiveness of current treatment choices. Therefore, the primary aim of this study was to describe cost-drivers of the top 1% and top ≥1-5% high-cost patients with sarcoidosis. The secondary aim was to compare costs of patients with and without fatigue complaints and to compare comorbidities.

Methods

Design and context

This study was a cross-sectional study using internal data of patients with sarcoidosis who visited the St. Antonius Hospital in the Netherlands. The study used administrative data provided by the business intelligence unit of the St. Antonius hospital. Patients were treated in the St. Antonius Hospital between January 1st 2011 and November 1st 2016. In total, 2251 patients with sarcoidosis were identified, from which 200 patients were used for the final analysis. When being enrolled in our cohort, there was at least of follow-up period of six months. The opening of the first diagnosis related group (DRG) due to their treatment for sarcoidosis was between the 11th of January 2011 and the 11th of May 2016.

We examined healthcare costs and identified the beneficiaries within the top 1% and the top ≥1%–5% of total costs. (16) The 2251 patients from our cohort were ranked in total costs after which the top 1% (0-0.99) and the ≥1-5% (1.0-4.99) most expensive patients were identified (n=110). From the remaining 2141 patients, a random sample of 90 patients was selected. Thus, from the total cohort, additional data was collected for 200 patients in total. Patients in the top 5% (so the top 1% and the top ≥1-5% together) of total health care costs were defined as high-cost patients.

Data collection

Baseline patient characteristics were examined for all patients with sarcoidosis (n=200). Patient characteristics included age, gender, BMI, Scadding stage, survival, fatigue, treatment time and the year of the diagnosis. Furthermore we collected information regarding the sarcoidosis related organ involvement (pulmonary, cardiac and neurologic). In addition, we collected data on whether or not patients suffered from pulmonary hypertension and obstructive sleep apnea (OSAS).

Mean treatment time was defined as the first date of receiving treatment minus the last date of receiving treatment at the St. Antonius hospital between January 1st 2011 and January 1st 2016. When the patient reported to experienced fatigue more than three times during the outpatient visits, he/she was seen as a patient with fatigue complaints in the analysis.

Healthcare utilization and cost calculation

Hospital administrative databases were extracted for all healthcare utilization as part of the diagnosis code for sarcoidosis patients. Next, this was grouped into six categories: 1) expensive medication (infliximab, adalimumab and rituximab), 2) intensive care/general ward nursing 3) respiratory medicine, 4) clinical chemistry, 5) radiology/ nuclear medicine, and 6) other.

Total costs per patient were calculated by summing the number of resources used multiplied by the costs per resource. We calculated costs for any given month in which care related to sarcoidosis was delivered in our hospital. Total annual costs were calculated by summing up the costs of all individual months the patient received care and subsequently correcting for the number of months. Months when no care was delivered were disregarded. Total costs per resource item were based on the national diagnosis treatment combination rates as defined in 2015 by the St. Antonius hospital. Costs of drugs were based on the lowest reported drug price according to the information of the Health Care Institute of the Netherlands valued in 2019. Total drug costs per patient were calculated using the amount of drugs (in mg) used multiplied with the price per mg in Euros.

Analyses

Descriptive statistics were used to present total costs. Costs were expressed as mean and interquartile range (IQR) due to skewness of the data. Overall mean costs (IQR) were presented for total health care costs made between January 2011 and January 2016. Treatment time and time between diagnosis/ first visit were expressed as mean (±SD). All categorical data were presented in *n* and percentage (%). All analyses were performed in SPSS (IBM SPSS Statistics version 24).

Results

For the 200 patients analysed, average total health care costs for the top 1%, top ≥1%–5% and bottom 95% beneficiaries were €108.296, €53.237 and €4.817, respectively. The mean annual cost are €51.082, €27.840 and €8.692, respectively. Mean treatment time in days for the top 1%, top ≥1-5% and the random sample was 1688 days (±225), 1412 days (±367) and 775 days (±659), respectively. Table 1 presents the demographics of the study population. In all groups, males were overrepresented, especially in the top 1%, where 68.2% of the patients were male. Mean age at diagnosis was comparable between the groups. In Table 2, the demographics are presented for the patients with less advanced sarcoidosis (without cardiac sarcoidosis, neurosarcoidosis or pulmonary hypertension). Here, top 1% beneficiaries were much older than the top ≥1-5% and the random sample of the bottom 95%. Furthermore, mortality was higher for high-cost patients, as 18% in the top 1% and 9.1% in the top ≥1%–5% died. In the random sample of the bottom 95%, 2.2% of the patients died.

Association of fatigue

In the top 1% and top ≥1%-5% 81.8% and 75% showed the experience fatigue complaints, respectively. In the random sample of the bottom 95%, 38.9% showed to have fatigue complaints.

In Table 3 the mean costs and patient characteristics are presented for patients with and without fatigue complaints for two groups: for the top 5% (the top 1% and top ≥1%-5% together) and for the random sample of the bottom 95%. Patients with and without fatigue showed to have comparable mean costs. Average total costs for the top 5% patients with fatigue were €65.512 and €60.165 without fatigue complaints. Average annual costs for the top 5% patients with fatigue were €33.039 and €30.311 without fatigue complaints. For the random sample of the bottom 95%, average total costs for patients with fatigue was €6.546 for patients with fatigue complaints and €3.716 for patients without fatigue complaints. In both the top 5% and random sample of the bottom 95%, patients with fatigue complaints were more likely to have pulmonary sarcoidosis, pulmonary hypertension, cardiac sarcoidosis, neurosarcoidosis and OSAS.

Table 1. Patient characteristics for three cost groups

Patient characteristics	Top 1%(n=22)	Top ≥1%-5%(n=88)	Sample of bottom 95%(n=90)
Male (n, %)	15 (68.2%)	52 (59.1%)	51 (56.7%)
Age at diagnosis (years ±SD)	44 ±12.3	43 ±11.0	45 ±12.6
Mean total costs (€, IQR)	€108.296 €25.497	€53.237 €39.384	€4.817 €4.466
Mean annual costs (€, IQR)	€51.082 €24.366	€27.840 €21.253	€8.692 €8.825
Treatment time (days ±SD)	1688 ±225	1412 ±367	775 ±659
Time between diagnosis/first visit (years ±SD)	8.8 ±6.2	7.3±6.7	6.1 ±9.1
BMI (±SD)	32.0 ±7.1	27.8 ±4.7	29.1 ±5.9
Fatigue (n, %)	18 (81.8%)	66 (75.0%)	35 (38.9%)
Deceased (n, %)	4 (18%)	8 (9.1%)	2 (2.2%)
Pul. sarc (n, %)	20 (90.9%)	81 (92.0%)	56 (62.2%)
Pul. Hypertension (n, %)	3 (13.6%)	9 (10.2%)	2 (1.7%)
Cardiac sarc (n, %)	2 (9.1%)	10 (11.3%)	5 (5.6%)
OSAS (n, %)	7 (31.8%)	9 (10.2%)	14 (15.6%)
Neurosarcoidosis (n, %)	6 (27.3%)	19 (21.6%)	12 (13.3%)
Scadding stage			
Scadding 0 (n, %)	4 (18.2%)	16 (18.2)	0 (0%)
Scadding I (n, %)	2 (9.1%)	13 (14.8%)	19 (21.1%)
Scadding II (n, %)	6 (27.3%)	18 (20.5%)	15 (16.7%)
Scadding III (n, %)	4 (18.0%)	13 (14.8%)	4 (3.0%)
Scadding IIII (n, %)	4 (18.0%)	28 (31.8%)	13 (14.4%)

IQR= Inter quartile range.

Comorbidities

In the top 1% and the top ≥1-5% there was more organ involvement of sarcoidosis compared to the random sample of the bottom 95%. Cardiac and pulmonary sarcoidosis and neurosarcoidosis were more likely in the top 1% and top ≥1-5%. OSAS was more present in the top 1% compared to the random sample of the bottom 95% (31.8% versus 15.6%). When patients with cardiac sarcoidosis, neurosarcoidosis or pulmonary hypertension were excluded, OSAS was still more present in the top 1% of the patients compared to both the top ≥1-5% and the random sample of the bottom 95% (33% versus 5% and 10%, respectively).

Cost-driver profile

The top 1% (n=22) spent a total of €3824 thousand. The top ≥1-5% patients (n=88) spent a total of €3,824,766. Figure 1A-C presents the share per category in the total costs. The top 3 cost-drivers identified in both the top 1% and top ≥1-5% high-cost patients were: 1) expensive medication, 2) intensive care/general ward nursing, and 3) costs made at the respiratory medicine department. Finally, the random sample of the bottom 95% spent a total of €442,019. Expenses made at the respiratory medicine department was the main cost-driver. Within the cost for expensive medication, infliximab accounted for 83% of the costs in the top 1%. In the

Table 2. Patient characteristics for three cost groups (without cardiac sarcoidosis, neurosarcoidosis and pulmonary hypertension)

Patient characteristics*	Top 1% (n=12)	Top ≥1%-5% (n=51)	Sample of bottom 95% (n=70)
Male (n, %)	8 (66.7%)	31 (60.8%)	39 (55.7%)
Age (years ±SD)	47 ±14.3	41± 11.0	44 ±12.1
Mean total costs (€, IQR)	€107.970	€50.861	€4.184
	€24.064	€45.551	€3.844
Mean annual costs (€, IQR)	€50.837	€25.322	€9.472
	€22.245	€17.413	€10.307
Treatment time (days ±SD)	1716 ±243	1411 ±352	700 ±650
Time between diagnosis/first visit (years ±SD)	7.9 ±7.8	7.3 ±5.7	5.9 ±9
BMI (±SD)	33.8 ±6.9	27.0 ±4.0	28.7 ±4.9
Fatigue (n, %)	10 (83.3%)	35 (68.6%)	21 (30%)
Deceased (n, %)	2 (16.7%)	5 (9.8%)	0 (0.0%)
Pul. sarc (n, %)	11 (91.7%)	47 (92.2%)	44 (62.9%)
OSAS (n, %)	4 (33.0%)	3 (5.0%)	7 (10.0%)
Scadding stage			
Scadding 0 (n, %)	2 (16.7%)	7 (13.7%)	5 (7.1%)
Scadding I (n, %)	0 (0.0%)	5 (9.8%)	14 (20.0%)
Scadding II (n, %)	3 (25.0%)	13 (25.5%)	15 (21.4%)
Scadding III (n, %)	3 (25.0%)	9 (17.6%)	4 (5.7%)
Scadding IIII (n, %)	4 (33.3%)	17 (33.3%)	9 (12.9%)
Unknown	0	0	23 (32.9%)

IQR= Inter quartile range. *without cardiac sarcoidosis, neurosarcoidosis and pulmonary hypertension.

top ≥1-5% this was 80% and in the random sample this was 98%. Adalimumab accounted for 16%, 19% and 0% of the total costs for expensive medication, respectively.

Discussion

In this study, we provide cost-related information and describe characteristics of 200 patients with sarcoidosis in a Dutch patient cohort using administrative data. The health care cost distribution for patients with sarcoidosis are highly skewed. The average healthcare costs of the top 1% patients with sarcoidosis were 22 times higher compared to that of the random sample of the bottom 95% (€108.296 vs. €4.817, respectively). The mean annual healthcare costs of the top 1% patients with sarcoidosis were 6 times higher compared to that of the random sample of the bottom 95% (€51.082 vs. €8.692, respectively). Treatment time of the most expensive 1% was 2.5 times longer compared to the random sample. The top 1% patients and top ≥1-5% patient showed to have more sarcoidosis related organ involvement compared to the random sample of the bottom 95%, indicating those are patients with more advanced sarcoidosis. Unexpectedly, patients with and without fatigue showed to have comparable mean costs and mean annual costs.

Although methodology and the data source used differ, our findings are consistent with other manuscripts studying the costs in patients with sarcoidosis. (12–14) Moreover, one study showed that patients in the top 5% in terms of costs, spent $93,201. (14) An-

Table 3. Patient characteristics for patients with and without fatigue

Patient characteristics	Top 0-5%		Random sample	
	With fatigue (n=84)	Without fatigue (n=26)	With fatigue (n=35)	Without fatigue (n=55)
Male (n, %)	49 (58.3%)	18 (69.2%)	15 (42.9%)	5 (9.1%)
Age (years ±SD)	53 (±11.1)	48 (±9.0)	51 (±12.5)	52 (±11.5)
Mean costs (€, IQR)	€65.512 (€44.486)	€60.165 (€54.307)	€6.546 (€6.080)	€3.716 (€2.552)
Mean annual costs (€, IQR)	€33.039 (€22.439)	€30.311 (€35.966)	€10.129 (€8.479)	€7.704 (€10.068)
Treatment time (days ±SD)	1484 (±347)	1410 (±401)	978 (±679)	634 (±613)
Time between diagnosis/first visit (years ±SD)	7.4 (±6.8)	8.3 (±5.7)	6.9 (±9.1)	5.6 (±9.0)
BMI (±SD)	28.7 (±5.3)	28.6 (±6.2)	29.7 (±6.8)	28.6 (±5.2)
Deceased (n, %)	10 (11.9%)	2 (7.7%)	1 (52.9%)	0 (0%)
Pulmonary sarcoidosis (n, %)	78 (92.9%)	23 (88.5%)	24 (68.6%)	32 (58.2%)
Pulmonary hypertension (n, %)	10 (11.9%)	2 (7.7%)	2 (5.7%)	1 (1.8%)
Cardiac sarcoidosis (n, %)	10 (11.9%)	2 (7.7%)	4 (11.4%)	6 (10.9%)
OSAS (n, %)	16 (19%)	0 (0%)	8 (22.9%)	0 (0%)
Neurosarcoidosis (n, %)	22 (26.2%)	3 (11.5%)	7 (20.0%)	5 (9.1%)
Scadding III (n, %)	13 (15.5%)	4 (15.4%)	2 (5.7%)	2 (3.6%)
Scadding IV (n, %)	23 (27.4%)	11 (42.3%)	7 (20.0%)	6 (10.9%)

IQR= Inter quartile range.

other study showed that the mean annual health care costs for patients in the top quintile were $73,346 (based on 2015 data). (13) This was 10 times greater compared to the mean annual health care costs for the remaining patients. Further, to our knowledge, this is the first study to use such data to characterize high-cost patients with sarcoidosis and identify key drivers that contribute to costs in this patient population.

This study has a number of limitations. First, we have studied a group of patients visiting our center for a period of 5 years. Thus, total costs for the full treatment time per patient are not presented in this study. Moreover, it would have been interesting to see total costs over a full treatment period per patient and next, to identify the most expensive treatment period looking at the patient cohort. Secondly, we were unable to include the societal costs. This includes costs due to work loss of patients not being able to work due to the severity of their disease course. Thirdly, this study was based on a population in the Netherlands. Therefore, this information cannot be generalized to other populations where cost of health care utilization can be very different. Also, some treatment options can be more expensive in other countries. It is known that performing a PET scan is more expensive in the USA compared to the Netherlands. As a consequence, the PET scan is performed less often in the USA. If we would leave out the PET scan cost, there would be no major changes in the cost distribution presented in Figure 1. Fourthly, we did not have a sufficient number of patients to perform an analysis of cost of the top 5% for the four major groups (neurosarcoidosis, cardiac sarcoidosis, pulmonary hypertension, and advanced sarcoidosis pulmonary). In a larger population it would have been very interesting to see whether total annual costs vary, depending upon underlying morbidity. Fifthly, we were unable to collect data when a patient was hospitalized outside our medical center.

Table 4. Patient characteristics for patients with and without fatigue (without cardiac sarcoidosis, neurosarcoidosis and pulmonary hypertension)

Patient characteristics	Top ≥1-5%		Random sample	
	With fatigue (n=45)	Without fatigue (n=18)	With fatigue (n=21)	Without fatigue (n=49)
Male (n, %)	27 (60%)	12 (66.7%	8 (38.1%%)	31 (63.3%)
Age (years ±SD)	52 (±12.1)	48 (±8.9)	47 (±11.9)	52 (±11.4)
Mean costs (€, IQR)	€65.044 (€46.618)	€53.476 (€48.151)	€6.103 (€3.361)	€3.361 (€5.632)
Mean annual costs (€, IQR)	€31.220 (€23.170)	€26.497 (€24.401)	€12.787 (€10.036)	€7.893 (€10.890)
Treatment time (days ±SD)	1502 (±326)	1387 (±414)	896 (±683)	609 (±621)
Time between diagnosis/first visit (years ±SD)	6.7 (±6.0)	9.1 (±6.1)	6.0 (±8.2)	5.9 (±9.4)
BMI (±SD)	28.9 (±5.3)	27.0 (±5.3)	28.5 (±4.6)	28.8 (±5.1)
Deceased (n, %)	5 (11.1%)	2 (11.1%)	0 (0%)	0 (0%)
Pulmonary sarcoidosis (n, %)	42 (93.3%)	16 (88.9%)	15 (71.4%)	29 (59.2%)
OSAS (n, %)	7 (15.6%)	0 (0%)	3 (14.3%)	4 (8.2%
Scadding III (n, %)	8 (17.8%)	4 (22.2%)	2 (9.5%)	2 (4.1%)
Scadding IV (n, %)	13 (28.9%)	8 (44.4%)	4 (19%)	5 (10.2%)

IQR= Inter quartile range. *without cardiac sarcoidosis, neurosarcoidosis and pulmonary hypertension

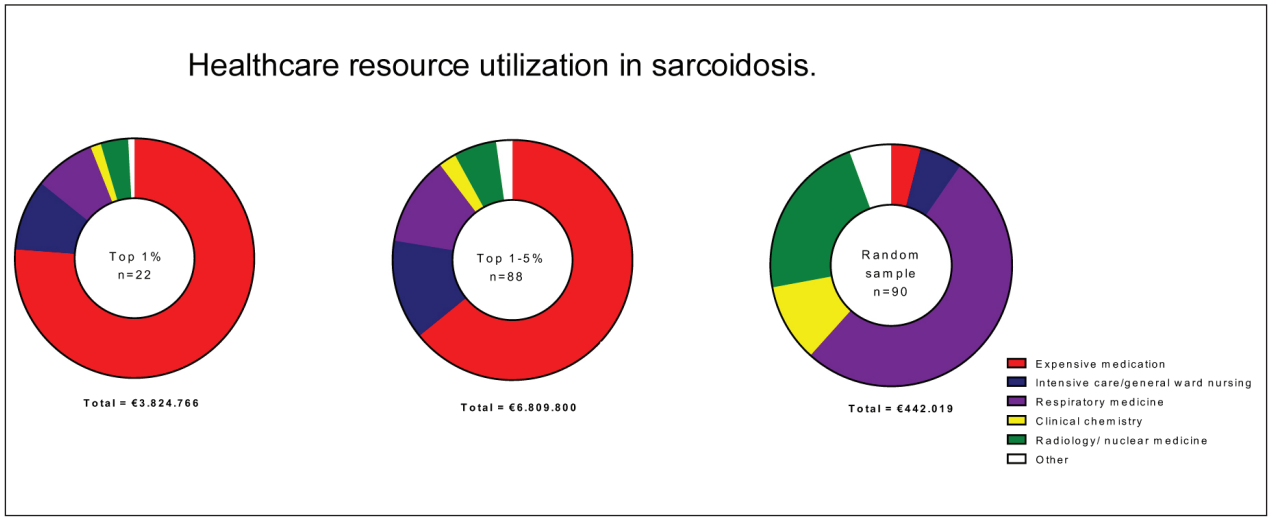

Fig. 1. Distribution of costs by resource categories for the top 1%, top ≥1-5% high-cost patients and the low-cost random sample.

However, we do not expect this to be a major (hidden) cost-driver. The Netherlands is a small country and planned admissions of tertiary patients are generally located in our center. A single emergency admission may sometimes take place regionally, but some of these patients will be transferred to us within a few days. Sixthly, we did not look into individual treatment schemes and were therefore unable to re-

port whether the patients were actively treated for e.g. pulmonary hypertension and how this affected their (medication) costs. Finally, a limitation with using annual costs is that we only used costs when care was delivered on a monthly basis. So when a patient would come back after six months for follow-up, the months in between visits were not part of our annual costs definition.

In conclusion, this study found that high-cost patients with sarcoidosis were patients with higher rates of comorbidities and had increased use of health care resources. Specifically, they were more often in need of expensive medication for their treatment. Both the management as well as physicians can specifically use this information to realize improvements in the quality of care and reducing overall costs for patients diagnosed with sarcoidosis, especially in referral centers of excellence. Efforts to further improve the quality of care and clinical outcomes for patients with sarcoidosis could specifically target the most expensive patients, which can potentially reduce the overall costs.

Support statement: This work was supported by The Netherlands Organisation for Health Research and Development (ZonMw) under project number 842001005. The funder had no role in the study design, data collection, analysis or decision of where to publish the manuscript.

References

1. Baughman RP, Lower EE, Gibson K. Pulmonary manifestations of sarcoidosis. Presse Med. Elsevier; 2012; 41: e289–e302.
2. Drent M, Lower EE, De Vries J. Sarcoidosis-associated fatigue. Eur. Respir; 2012; 40: 255–263.
3. Costabel U, Hunninghake GW, Committee SS. ATS/ERS/WASOG statement on sarcoidosis. Eur. Respir. J. Wiley Online Library; 1999; 14: 735–737.
4. Iannuzzi MC, Fontana JR. Sarcoidosis: clinical presentation, immunopathogenesis, and therapeutics. Jama American Medical Association; 2011; 305: 391–399.
5. De Vries J, Drent M. Quality of life and health status in sarcoidosis: a review. Semin. Respir. Crit. Care Med. New York: Thieme Medical Publishers, c1994-; 2007. p. 121–127.
6. Cox CE, Donohue JF, Brown CD, Kataria YP, Judson MA. Health-related quality of life of persons with sarcoidosis. CHEST J. American College of Chest Physicians; 2004; 125: 997–1004.
7. Michielsen HJ, Drent M, Peros-Golubicic T, De Vries J. Fatigue is associated with quality of life in sarcoidosis patients. CHEST J. American College of Chest Physicians; 2006; 130: 989–994.
8. de Kleijn WP, De Vries J, Lower EE, Elfferich MD, Baughman RP, Drent M. Fatigue in sarcoidosis: a systematic review. Curr. Opin. Pulm. Med. CoRPS, Department of Medical Psychology, Tilburg University, Tilburg, the Netherlands.; 2009; 15: 499–506.
9. Marcellis RG, Lenssen AF, Elfferich MD, De Vries J, Kassim S, Foerster K, Drent M. Exercise capacity, muscle strength and fatigue in sarcoidosis. Eur. Respir. J. Dept of Respiratory Medicine, ild Care Consultancy, Maastricht University Medical Centre, NC Maastricht, The Netherlands.; 2011; 38: 628–634.
10. Hinz A, Fleischer M, Brähler E, Wirtz H, Bosse-Henck A. Fatigue in patients with sarcoidosis, compared with the general population. Gen. Hosp. Psychiatry Elsevier; 2011; 33: 462–468.
11. Blumenthal D, Abrams MK. Tailoring complex care management for high-need, high-cost patients. Jama American Medical Association; 2016; 316: 1657–1658.
12. Rice JB, White A, Lopez A, Conway A, Wagh A, Nelson WW, Philbin M, Wan GJ. Economic burden of sarcoidosis in a commercially-insured population in the United States. J. Med. Econ. Taylor & Francis; 2017; 20: 1048–1055.
13. Rice JB, White A, Lopez A, Nelson WW. High-Cost Sarcoidosis Patients in the United States: Patient Characteristics and Patterns of Health Care Resource Utilization. J. Manag. care Spec. Pharm. Academy of Managed Care Pharmacy; 2017; 23: 1261–1269.
14. Baughman RP, Field S, Costabel U, Crystal RG, Culver DA, Drent M, Judson MA, Wolff G. Sarcoidosis in America. Analysis based on health care use. Ann. Am. Thorac. Soc. Am Thoracic Soc; 2016; 13: 1244–1252.
15. Kaplan RS, Porter ME. How to solve the cost crisis in health care. Harv. Bus. Rev. Harvard Business School, USA.; 2011; 89: 46–52, 54, 56-61 passim.
16. Wammes JJG, Tanke M, Jonkers W, Westert GP, Van der Wees P, Jeurissen PP. Characteristics and healthcare utilisation patterns of high-cost beneficiaries in the Netherlands: a cross-sectional claims database study. BMJ Open; 2017; 7: e017775-2017–017775.