

INCIDENCE AND ECONOMIC BURDEN OF SARCOIDOSIS IN YEARS 2011-2015 IN SILESIAN VOIVODESHIP, POLAND

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ABSTRACT. *Background:* Sarcoidosis is a rare, chronic systemic disease. Earlier data (2006-2010) suggest that the incidence of pulmonary sarcoidosis in Silesian voivodeship increased, however there is no current data on other clinical forms of the disease. *Objectives:* The aim of presented study was an analysis of the actual epidemiological situation of sarcoidosis with simultaneous estimation of treatment cost financed from public funds. *Methods:* Epidemiological descriptive study concerned registered cases of sarcoidosis diagnosed in adult inhabitants of the Silesian voivodeship in years 2011-2015. Secondary epidemiological data on the main diagnosis and comorbidity were obtained from the National Health Fund (NFZ) database in Katowice. Territorial and temporal variability of standardized incidence rates were analysed with simultaneous estimation of treatment costs reimbursed from the state budget. *Results:* Pulmonary sarcoidosis was the most frequently registered clinical form of such disease in the Silesian voivodeship (65% of total cases). The highest number of cases was diagnosed in the age 35-54 years, frequently in men than in women. Significantly decrease of the standardized incidence of sarcoidosis noticed between 2011 and 2015 is related with observed lower number of total cases of pulmonary disease. Observed territorial variability of the sarcoidosis incidence requires future, well-planned studies. The annual average direct cost of sarcoidosis treatment is high and exceed 538 EUR per patient. *Conclusions:* It was confirmed that sarcoidosis in the Silesian Voivodeship is a rare disease, however reimbursed direct costs of treatment remains very high. (*Sarcoidosis Vasc Diffuse Lung Dis* 2020; 37 (1): 43-52)

KEY WORDS: sarcoidosis, incidence, economic burden, descriptive study

INTRODUCTION

Sarcoidosis is a chronic disease manifested by the presence of granulomas in many tissues or organs, mainly in lungs, but also in lymph nodes, heart, organs of sight, liver or heart (1-3, 8, 9). The aetiology of the disease remains unexplained (4) although there are reports on genetic determinants appropri-

ate for specific ethnic groups (5-7, 9, 12, 13), and also indicating an abnormal immune response due to exposure (6, 9, 12, 13). The factors include: fungi, mycobacteria, bacteria, parasites (1, 6, 13) and pollen, metals, chemicals, or medicines (1, 9, 13).

The incidence of sarcoidosis substantially differs between regions, the highest values were reported in the African-American population of the United States (35.5/100,000), as well as in the populations of Northern European countries (Sweden 24/100,000, Norway 14-15/100,000, Finland 11.4/100,000) (6, 10). The lowest incidence was recorded in Southern European countries (Spain 0.42/100,000, Greece 1.07/100,000) and also in Japan (0.56-1.01/100,000 population) (10). The incidence of pulmonary sar-

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coidosis in Poland, estimated in years 2006–2010 was at the level of 3.8–4.5/100,000 population (11).

There is no separate registry of particular form of sarcoidosis hospitalization cases in Poland, current registry include the number of entire cases of certain disorders involving the immune mechanism (D86) with pulmonary sarcoidosis and other allergic lung diseases, as well as autoimmune and granulomas (14). In the period 2011–2015, the number of hospitalizations due to total cases of diseases in this group was constantly increasing and ranged from 6,589 to 8,113 per year.

The cost of sarcoidosis treatment is high, eg. total annual direct cost of treatment in the United States is about 1.3–8.7 billion USD, and the average annual cost is at the level USD 19,714 per patient (15). Moreover, it is documented that the largest annual average cost of treatment of a patient with sarcoidosis (80–100th percentile of total costs) reached USD 7,345 (EUR 59,719.76) (16). The average cost of total granuloma treatment in the period 2011–2015, in Poland are stable and ranged PLN 4,850.18 (EUR 1,149.99) per patient and PLN 4,943.76 (EUR 1,172.17), in 2011 and 2013 year respectively (14).

Observed progressive aging of population causes increase the number of chronic diseases, including number of patients hospitalized due to sarcoidosis (11, 14). Those observation justify needs of epidemiological study in the aim of current situation assessment in one of the biggest Polish agglomeration (Silesian Voivodeship), including incidence, hospitalization and total cost of treatment granted from the government funds.

METHODS

This paper presents results of descriptive epidemiological study conducted in the Silesian voivodeship and based on secondary epidemiological data registered by National Health Fund (NHF) in Katowice, in years 2011–2015. Obtained data on sarcoidosis (D86; ICD-10 version) include addresses of services providers, type of service (outpatient, stationary), admission mode, mode of discharge and cost of benefits. Moreover, anonymous patient data were collected, age, gender and place of residence, and the major diagnosis code according to ICD-10

along with three comorbidities. The including criterion was ever diagnosed sarcoidosis recognized as the main diagnosis and/or as one of the comorbidities. The following forms of the disease were included in final database: sarcoidosis of the lung (D86.0) and sarcoidosis of the lung with sarcoidosis of lymph nodes (D86.2), sarcoidosis of lymph nodes (D86.1), sarcoidosis of skin (D86.3), sarcoidosis of other sites (D86.8) and unspecified sarcoidosis (D86.9).

The number and percentage of patients with sarcoidosis, separately women and men in the following age groups: 19–34, 35–54, 55–64 and 65+, were determined. Crude and standardized incidences of sarcoidosis in particular years of the study period (2011–2015) were presented as a rate per 100,000 population of 19+ years. The average number of inhabitants in 2011–2015 was 3,722,496 ($3,712,784 \div 3,728,366$). Their temporal variability in the Silesian Voivodeship was shown in separated local administrative units according to NTS-4 (Nomenclature of Territorial Units for Statistics), for which detailed description of procedure was presented in the earlier publication (11). The assessment of sarcoidosis territorial variability was presented on contour maps of the Silesian Voivodeship as an averaged value of incidence rates in 2011–2015. For this purpose, the geographical information system ArcGIS 9.2 was used. Moreover, the direct costs of sarcoidosis treatment, in both outpatient visits and hospitalization, incurred in 2011–2015 were estimated. On the basis of data on the services number in particular reported years, the average unit costs of disease treatment in PLN were calculated. The structure of these expenses was presented based on the following rounded quantile values: $k_{0.25}=50$, $k_{0.5}=500$, $k_{0.75}=5,000$. The study wasn't any medical experiment, and the secondary character of data didn't need Bioethics Committee permission.

Statistical analysis of data was based on MS Excel 2013 (Microsoft Office 2013) software and the 2.11.1 R package (GNU GPL license).

RESULTS

According to the assumed aim of the study, appropriate crude and standardized incidence rates were calculated, detailed results are presented in Table 1. As it was expected, the standardized values were almost two times smaller than the crude val-

ues: 7.08-13.08/100,000 vs 12.04-22.09/100,000 population 19+ years. The highest standardized in-

cidence rate was obtained for pulmonary sarcoidosis with values ranging from 9.55/100,000 in 2011 to

Table 1. Crude and standardized incidence rates of sarcoidosis (D86; ICD-10 version) in adults aged 19 and over (n/100,000), Silesian Voivodeship

Year	Main diagnosis				Main or co-existing diagnosis			
	Crude ratio	Standardized ratio			Crude ratio	Standardized ratio		
		Total	Total	Women		Men	Total	Total
Sarcoidosis D86								
2011	22.09	13.08	11.31	14.71	25.65	15.02	12.93	17.00
2012	18.27	10.86	11.01	10.45	21.32	12.57	12.73	12.13
2013	13.80	8.39	7.40	9.31	16.22	9.80	8.64	10.85
2014	12.28	7.71	6.16	9.20	14.41	8.91	7.16	10.60
2015	12.04	7.08	5.61	8.52	14.44	8.40	6.61	10.13
Sarcoidosis of lung D86.0, D86.2								
2011	16.07	9.55	7.69	11.32	18.36	10.78	8.56	12.93
2012	12.77	7.62	7.48	7.57	14.32	8.49	8.38	8.42
2013	8.86	5.33	4.69	5.94	10.34	6.18	5.39	6.92
2014	6.96	4.35	3.23	5.45	8.09	4.97	3.71	6.20
2015	7.35	4.32	3.47	5.16	8.65	5.05	3.89	6.18
Sarcoidosis of lymph nodes D86.1								
2011	2.68	1.57	1.51	1.61	2.90	1.69	1.61	1.77
2012	2.90	1.72	1.82	1.60	3.25	1.91	2.00	1.77
2013	2.26	1.43	1.29	1.55	2.52	1.60	1.41	1.76
2014	2.82	1.81	1.59	2.00	3.20	2.05	1.75	2.35
2015	2.50	1.56	1.16	1.95	2.91	1.76	1.35	2.16
Sarcoidosis of skin D86.3								
2011	0.27	0.15	0.30	0.00	0.35	0.20	0.39	0.00
2012	0.24	0.15	0.21	0.07	0.32	0.20	0.29	0.11
2013	0.35	0.19	0.16	0.22	0.43	0.24	0.22	0.26
2014	0.32	0.18	0.26	0.11	0.35	0.19	0.27	0.11
2015	0.13	0.05	0.07	0.02	0.16	0.07	0.12	0.02
Sarcoidosis of other sites D86.8								
2011	1.18	0.71	0.77	0.64	1.40	0.85	0.91	0.78
2012	0.67	0.37	0.51	0.23	0.83	0.45	0.56	0.34
2013	0.67	0.41	0.51	0.29	0.89	0.53	0.64	0.42
2014	0.51	0.33	0.34	0.32	0.56	0.36	0.39	0.32
2015	0.27	0.15	0.19	0.11	0.30	0.16	0.19	0.14
Unspecified sarcoidosis D86.9								
2011	1.88	1.09	1.03	1.14	2.66	1.50	1.46	1.52
2012	1.69	1.00	0.99	0.98	2.60	1.52	1.50	1.49
2013	1.66	1.04	0.75	1.31	2.04	1.25	0.99	1.49
2014	1.67	1.03	0.74	1.33	2.20	1.34	1.04	1.63
2015	1.78	1.00	0.72	1.28	2.42	1.36	1.07	1.64

4.32/100,000 in 2015 year. Moreover, the value was higher in men than in women (5.16-11.32/100,000 vs 3.23-7.69/100,000 respectively).

Figure 1 illustrates the territorial variability of sarcoidosis incidence rates (values averaged in the study period 2011-2015). The highest values were observed in the following cities: Siemianowice Śląskie, Gliwice, Ruda Śląska, Dąbrowa Górnicza and Tychy and districts: gliwicki, lubliniecki, bierunsko-ledziński. The lowest values concerned cities: Bytom, Zory and districts: wodzisławski, rybnicki, cieszyński.

It is worth noting that most frequently sarcoidosis was detected in people aged 35-54 years (Figure 2). Patients with sarcoidosis as the major diagnosis were somewhat younger (46.6 ± 13.3 years) than those with sarcoidosis as a co-occurring disease (47.3 ± 13.5 years).

Sarcoidosis mostly affected men than women (52.0% vs. 48%), both total and particular forms of the disease (Figure 3).

Detailed data on the number of new outpatient visits or hospitalization due to sarcoidosis was shown

in Table 2. It was noted, that a total number of cases and a number of outpatient visits significantly decreased in subsequent reporting years with a relatively stable number of hospitalizations in the entire study period 2011-2015. The highest number of patients was observed in the beginning year of the study.

Most frequently diagnosed form of sarcoidosis was the pulmonary form of the disease (D86.0, D86.2; ICD-10 version) and contained about 65% of all cases (N=3427) recognized in the study period (2012-2015) and over 70% of cases in the beginning year (N=956; 2011). Sarcoidosis of lymph nodes (D86.1) occurred in about 16% of patients, lower percentages were related to diagnoses of unspecified sarcoidosis (D86.9) - 10% of cases, sarcoidosis of other sites (D86.8) - 4% of cases, as well as sarcoidosis of skin (D86.3) - 2% of cases, in 2012-2015 (N=3427).

Table 3 presents a history of treating patients with sarcoidosis. In the case of over 80% of people, treatment started in a planned mode, based on a referral. Over half of patients with sarcoidosis (60%)

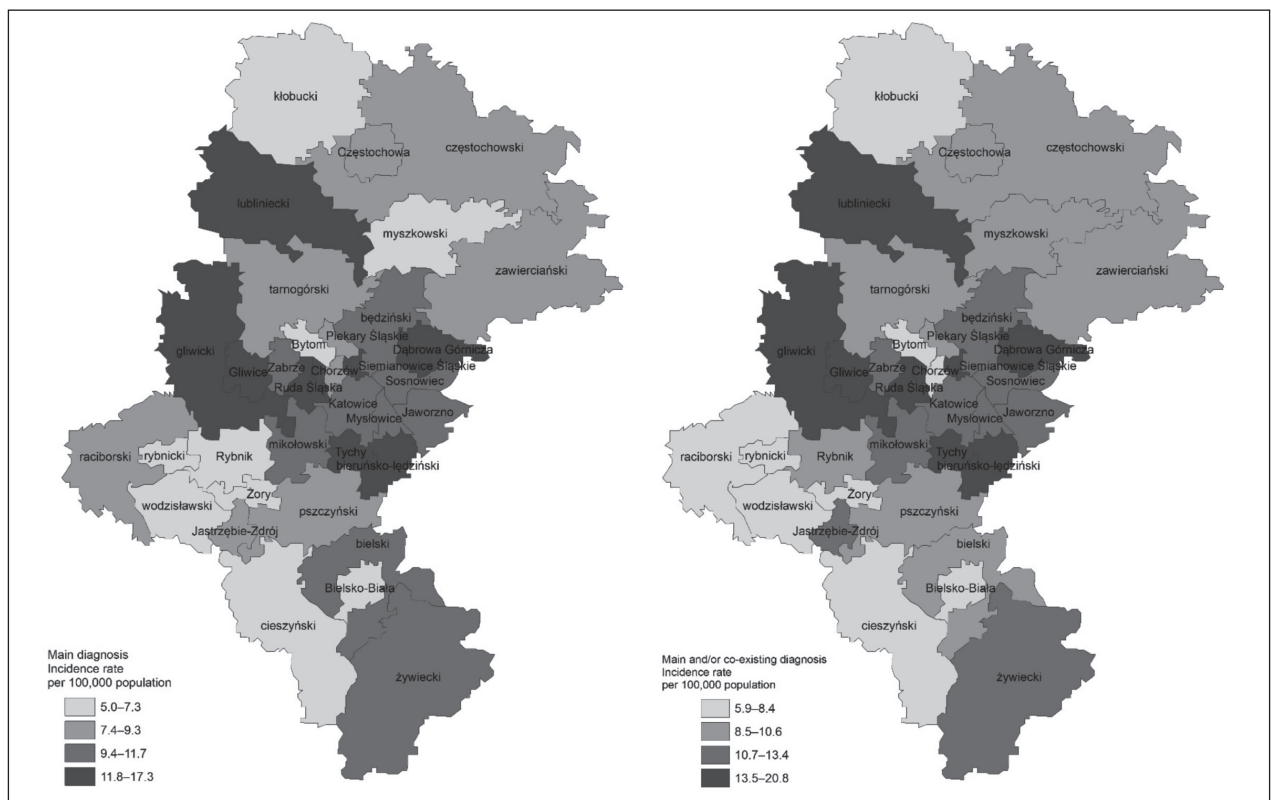


Fig. 1. Standardized incidence rates of sarcoidosis in adults inhabitants of Silesian voivodeship, values averaged for the period 2011-2015 (n/100,000)

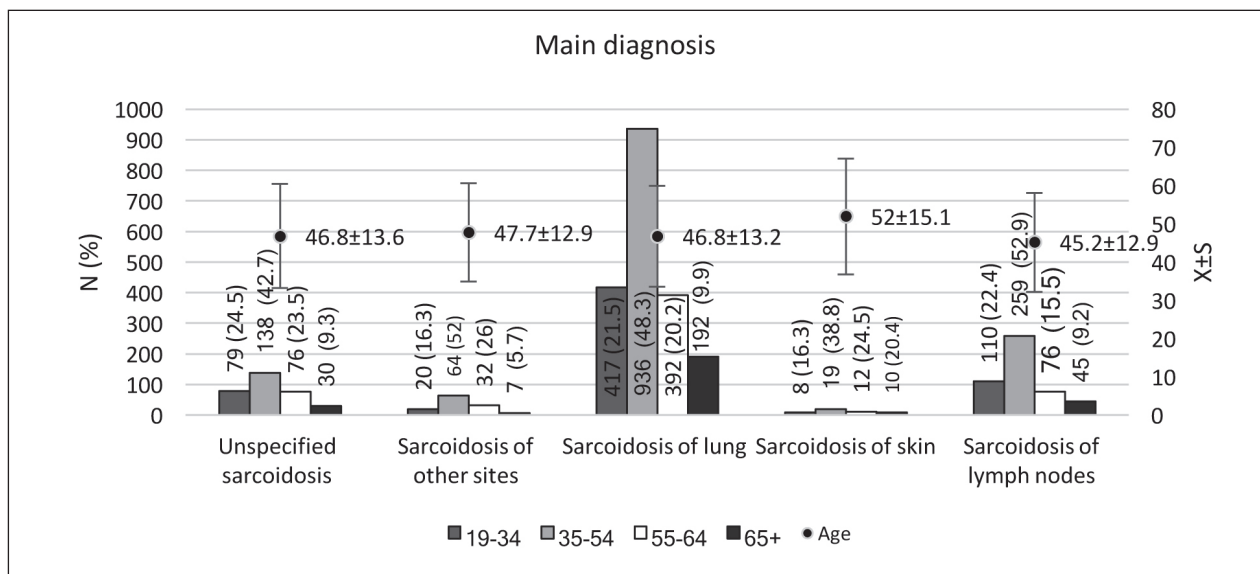


Fig. 2. Numbers and % of first-time diagnosed sarcoidosis (D86; ICD-10 version) in the entire period (2011-2015) by age of patients, Silesian Voivodeship

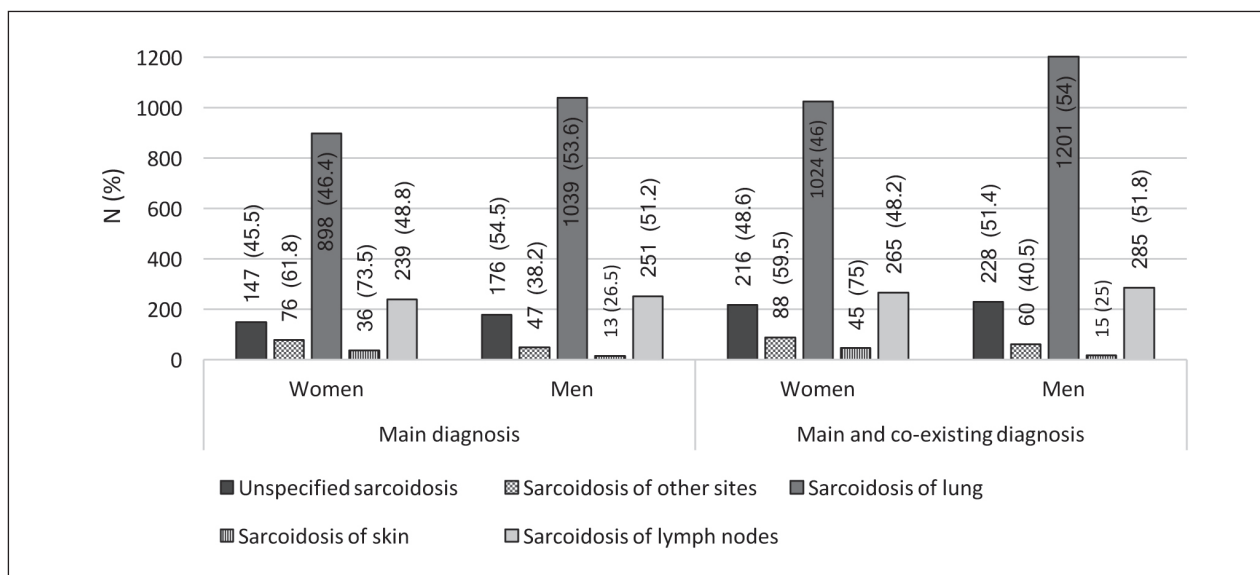


Fig. 3. Total number and percentage of first-time diagnosed sarcoidosis (D86; ICD-10 version) in the entire study period (2011-2015), Silesian Voivodeship

were referred for further treatment, after discharge from the hospital. In the study period, none patient died due to sarcoidosis as the major diagnosis.

The average costs of hospitalization patients with sarcoidosis as a major diagnosis (funds granted from the budget) were PLN 5,115.17 (EUR 1,177.80) per patient and were similar to those refunded in patients with sarcoidosis as a co-morbid

disease (PLN 4,684.03 - EUR 1 078.52). Detailed data are presented in Table 4.

It is worth noting that in the case of nearly 30% of unit services, the annual cost did not exceed PLN 50 per patient. The percentage of patients for which the funds spent were greater than PLN 5,000 did not exceed 13.8% (patients with the main diagnosis) and 12.5% (patients with comorbid sarcoidosis).

Table 2. The number of first-time outpatient visits or hospitalizations due to particular form of sarcoidosis in adults 19+ years, Silesian Voivodeship

Year	Total number of sarcoioidosis D86			Sarcoidosis of lung D86.0, D86.2			Sarcoidosis of lymph nodes D86.1			Sarcoidosis of skin D86.3			Sarcoidosis of other sites D86.8			Unspecified sarcoidosis D86.9			Number of total death
	O	H	Total N (%)	O	H	Total N (%)	O	H	Total N (%)	O	H	Total N (%)	O	H	Total N (%)	O	H	Total N (%)	
Main diagnosis																			
2011	425	398	823 (100)	305	294	599 (72.8)	51	49	100 (12.2)	8	2	10 (1.2)	25	19	44 (5.3)	36	34	70 (8.5)	5 (0.6)
2012	464	217	681 (100)	349	127	476 (69.9)	52	56	108 (15.9)	7	2	9 (1.3)	14	11	25 (3.7)	42	21	63 (9.3)	6 (0.9)
2013	298	216	514 (100)	210	120	330 (64.2)	34	50	84 (16.3)	7	6	13 (2.5)	10	15	25 (4.9)	37	25	62 (12.1)	16 (3.1)
2014	221	236	457 (100)	137	122	259 (56.7)	42	63	105 (23)	5	7	12 (2.6)	9	10	19 (4.2)	28	34	62 (13.6)	11 (2.4)
2015	201	246	447 (100)	132	141	273 (61.1)	36	57	93 (20.8)	2	3	5 (1.1)	3	7	10 (2.2)	28	38	66 (14.8)	27 (6)
Total N (%)	1609 (55.1)	1313 (44.9)	2922 (100)	1133 (58.5)	804 (41.5)	1937 (66.3)	215 (43.9)	275 (56.1)	490 (16.8)	29 (59.2)	20 (40.8)	49 (1.7)	61 (49.6)	62 (50.4)	123 (4.2)	171 (52.9)	152 (47.1)	323 (11.1)	65 (2.2)
Sarcoidosis as a main or co-existing diagnosis																			
2011	451	505	956 (100)	327	357	684 (71.5)	52	56	108 (11.3)	8	5	13 (1.4)	27	25	52 (5.4)	37	62	99 (10.4)	12 (1.3)
2012	491	304	795 (100)	369	165	534 (67.2)	54	67	121 (15.2)	8	4	12 (1.5)	15	16	31 (3.9)	45	52	97 (12.2)	12 (1.5)
2013	325	279	604 (100)	227	158	385 (63.7)	35	59	94 (15.6)	9	7	16 (2.6)	12	21	33 (5.5)	42	34	76 (12.6)	27 (4.5)
2014	246	290	536 (100)	149	152	301 (56.2)	49	70	119 (22.2)	5	8	13 (2.4)	10	11	21 (3.9)	33	49	82 (15.3)	22 (4.1)
2015	237	299	536 (100)	154	167	321 (59.9)	40	68	108 (20.1)	2	4	6 (1.1)	4	7	11 (2.1)	37	53	90 (16.8)	37 (6.9)
Total N (%)	1750 (51.1)	1677 (48.9)	3427 (100)	1226 (55.1)	999 (44.9)	2225 (64.9)	230 (41.8)	320 (58.2)	550 (16)	32 (53.3)	28 (46.7)	60 (1.8)	68 (45.9)	80 (54.1)	148 (4.3)	194 (43.7)	250 (56.3)	444 (13)	110 (3.2)

O - outpatient treatment visits; H - hospitalization, stationary treatment

Table 3. History of treatment patients with sarcoidosis in Silesian voivodeship (D86; ICD-10 version) entire study period (2011-2015)

Admission	Sarcoidosis as a major diagnosis N (%)	Sarcoidosis as a major or coexisting diagnosis N (%)
Outpatient treatment		
Emergency admission	157 (9.8)	171 (9.8)
Planned admission based on referral	1405 (87.3)	1498 (85.6)
No data	47 (2.9)	81 (4.6)
Total	1609 (100)	1750 (100)
Hospitalization (stationary treatment)		
Emergency admission	239 (18.2)	418 (24.9)
Planned admission based on a referral	1074 (81.8)	1259 (75.1)
Total	1313 (100)	1677 (100)
Discharge		
Referral for further treatment	799 (60.9)	1080 (64.4)
Discharge against medical advice (AMA)	7 (0.5)	10 (0.6)
The end of the therapeutic or diagnostic process	507 (38.6)	579 (34.5)
Death of the patient	0 (0)	8 (0.5)
Total	1313 (100)	1677 (100)

DISCUSSION

Obtained results confirmed that the current epidemiological situation of sarcoidosis (2011-2015) in the Silesian Voivodeship is satisfactory. First, we observed a decrease of new cases of disease which was basically related to decreasing the number of dominant clinical forms of the disease - pulmonary sarcoidosis (about 65% of cases). Similarly, lung sarcoidosis is the most common form of the disease in the USA, data of the Foundation for Sarcoidosis Research (FSR) suggests that this form of disease affected even up to 90% of patients (17). Extrapulmonary sarcoidosis includes the following forms: peripheral lymph nodes, heart, eyeballs, nervous system, skin, liver and/or spleen, the osteoarticular system (1, 12, 17). Own data confirmed that 16% of patients had sarcoidosis of lymph nodes, sarcoidosis of other sites occurred in 4% of patients, sarcoidosis of skin was diagnosed in 2%, and unspecified sarcoidosis in more than 10%. A similar structure of disease was observed in the Netherlands where pulmonary form concerned 82% of patients, ocular sarcoidosis 3.8%, neurosarcoidosis 3.3%, cardiac sarcoidosis 1.6%, hypercalcemia 2.2% and sarcoidosis of skin and other symptoms occurred in 7.1% of patients (18). The

Case-Control Etiologic of Sarcoidosis Study (ACCESS) indicated, that the dominant form was the pulmonary sarcoidosis (51.9% of patients), next sarcoidosis of skin (12.7%), sarcoidosis of other sites (6.3%) and unspecified sarcoidosis (15.9%) (25).

Treatment of sarcoidosis has an individual character without unambiguous standards methods (19). The Polish NHF registry indicated that more than half of patients (55.1%) in the Silesian Voivodeship were treated in out-patient visits immediately after the first-time diagnosis, whereas 1313 (44.9%) of patients were hospitalized. Most people with pulmonary sarcoidosis and sarcoidosis of skin were patients of outpatient treatment, while people with sarcoidosis of lymph node and sarcoidosis of other sites rather required hospitalization. The observed difference was most likely a consequence of better availability of detailed diagnostic methods and appropriate therapy in hospitals only (2). The results of other studies in the United States revealed that the rate of hospitalization due to sarcoidosis was significantly higher than the rates of hospitalization of patients without sarcoidosis (17.3/100 vs 12.6/100 person-years, respectively) (22). We observed, that 60% of patients hospitalized due to first-time diagnosed sarcoidosis in the Silesian voivodeship were referred for further treatment.

Table 4. Total and unit cost of health services granted from the budget (PLN) in particular form of sarcoidosis. (PLN to EUR according to the NBP exchange rate from 2018-06-28: EUR/PLN = 0.2303)

Diagnosis	Actual direct costs of treatment patients with sarcoidosis PLN / EUR					Total cost 2011-2015 PLN / EUR	Average unit cost 2011-2015 PLN / EUR	Total cost 2011-2015 PLN / EUR		Average unit cost 2011-2015 PLN / EUR		
	2011	2012	2013	2014	2015			O	H	O	H	
Main diagnosis	Unspecified sarcoidosis	152,188.50	101,237.10	123,520.85	172,780.65	171,454.25	721,181.35	2,332.76	11,960.35	709,221.00	69.94	4,665.93
		35,042.25	23,310.41	28,441.36	39,783.71	39,478.30	166,056.02	514.11	2,753.94	163,302.09	16.10	1,074.36
	Sarcoidosis of other sites	93,654.75	58,557.00	81,037.00	49,249.50	30,469.00	312,967.25	2,544.45	3,173.75	309,793.50	52.03	4,996.67
		21,564.53	13,483.08	18,659.22	11,339.97	7,015.66	72,062.45	585.87	730.77	71,331.68	11.98	1,150.51
	Sarcoidosis of lung	1,615,996.55	708,757.16	635,279.51	665,461.36	666,388.90	4,291,883.48	2,215.74	82,581.95	4,209,301.53	72.89	5,235.45
		372,092.21	163,195.28	146,276.65	153,226.19	153,439.76	988,230.08	510.19	19,014.95	969,215.13	16.78	1,205.49
	Sarcoidosis of skin	3,648.53	3,620.90	11,348.15	14,137.90	5,248.45	38,003.93	775.59	1,064.93	36,939.00	36.73	1,846.95
		840.09	833.73	2,612.97	3,255.33	1,208.48	8,750.62	178.58	245.21	8,505.41	8.46	425.27
	Sarcoidosis of lymph nodes	257,533.65	329,574.10	288,272.20	332,850.22	257,750.79	1,465,980.96	2,991.80	15,012.89	1,450,968.07	69.83	5,276.25
		59,298.56	75,886.27	66,376.28	76,640.62	59,348.56	337,550.28	688.88	3,456.80	334,093.48	16.08	1,214.89
Total	2,123,021.98	1,201,746.26	1,139,457.71	1,234,479.63	1,131,311.39	6,830,016.97	2,337.45	113,793.87	6,716,223.10	70.72	5,115.17	
	488,837.64	276,708.77	262,366.49	284,245.81	260,490.75	1,572,649.46	538.21	26,201.67	1,546,447.79	16.28	1,177.80	
The average unit cost per patient	2,579.61	1,764.68	2,216.84	2,701.27	2,530.90	2,337.45	-	70.72	5,115.17	-	-	-
	593.97	406.33	510.44	621.98	582.75	538.21	-	16.28	1,177.80	-	-	-
Main or co-existing diagnosis	Unspecified sarcoidosis	243,287.95	190,041.31	145,769.45	243,701.19	234,680.25	1,057,480.15	2,381.71	12,871.55	1,044,608.60	66.35	4,178.43
		56,018.41	43,758.07	33,564.23	56,113.56	54,036.44	243,490.70	548.40	2,963.75	240,526.95	15.28	962.11
	Sarcoidosis of other sites	103,873.95	67,100.60	109,878.74	51,186.60	30,507.80	362,547.69	2,449.65	3,547.59	359,000.10	52.17	4,487.50
		23,917.56	15,450.29	25,300.19	11,786.00	7,024.59	83,478.63	564.05	816.85	82,661.77	12.01	1,033.27
	Sarcoidosis of lung	1,786,760.77	823,568.31	759,642.43	746,828.05	749,027.59	4,865,827.14	2,186.89	86,678.28	4,779,148.87	70.70	4,783.93
		411,411.62	189,631.19	174,911.90	171,961.32	172,467.78	1,120,383.81	503.54	19,958.16	1,100,425.65	16.28	1,101.53
	Sarcoidosis of skin	8,426.53	6,457.60	12,779.45	15,820.90	8,368.45	51,852.93	864.22	1,120.93	50,732.00	35.03	1,811.86
		1,940.26	1,486.90	2,942.54	3,642.85	1,926.88	11,939.43	198.99	258.10	11,681.33	8.07	417.19
	Sarcoidosis of lymph nodes	286,728.45	384,184.30	327,536.67	350,402.22	288,338.94	1,637,190.58	2,976.71	15,554.24	1,621,636.34	67.63	5,067.61
		66,020.82	88,460.58	75,417.14	80,682.06	66,391.65	376,972.25	685.40	3,581.45	373,390.80	15.57	1,166.85
Total	2,429,077.65	1,471,352.12	1,355,606.74	1,407,938.96	1,310,923.03	7,974,898.49	2,327.08	119,772.59	7,855,125.90	68.44	4,684.03	
	559,308.66	338,787.02	312,136.00	324,185.79	301,847.33	1,836,264.81	535.62	27,578.31	1,808,686.50	15.76	1,078.52	
The average unit cost per patient	2,540.88	1,850.76	2,244.38	2,626.75	2,445.75	2,327.08	-	68.44	4,684.03	-	-	-
	585.05	426.15	516.78	604.82	563.15	535.82	-	15.76	1,078.52	-	-	-

O - outpatient treatment; H - hospitalization, stationary treatment

The demographic structure of patients with pulmonary sarcoidosis in the present study was similar to those observed in earlier research period 2006–2010 (11). The frequency of disease was the highest in people aged 35–54 years. Polish and USA studies confirmed that sex is a serious determinant of the diagnosed form of the disease, sarcoidosis of lymph node and unspecified sarcoidosis were dominant forms in Polish males while in female sarcoidosis of skin and sarcoidosis of other sites were the major forms (11, 20). In the USA neurosarcoidosis, ocular sarcoidosis and erythema nodular were significantly more frequently recognized in women than in men (20).

Obtained results confirmed significant temporal variability of sarcoidosis incidence in the Silesian voivodeship. During an earlier period (years 2006–2010) an increase of the standardized incidence rate was observed from the value of 3.8/100,000 to 4.3/100,000. Next in 2011 was noted a double increase of incidence to the value 9.55/100,000 population. Current data confirmed a significant decrease in incidence rate in years 2012–2015 to the values 7.62–4.32/100,000 population respectively. Such observation is difficult to explaining in the case of descriptive study thus, it cannot be ruled out that an unstable register is responsible for this variability, and we have to project future research in which confounder and modifying factors will be taken into account. However, contrary to our observations, the epidemiological situation of sarcoidosis in the United States was quite stable, the incidence in the years 2010–2013 was at the level 7.6–8.8/100,000 population. Similarly, stable values of crude incidence in the years 1991–2003 was recorded in Great Britain, the incidence of sarcoidosis was in the range of 4.45–5.59/100,000 population (23). Concluding, the current incidence of sarcoidosis in Silesian Voivodeship is consistent with some other cited studies. Significantly higher values apply to Northern European countries with values 24/100,000 in Sweden, 11.4/100,000 in Finland, 14–15/100,000 in Norway and 7.2/100,000 in Denmark. On the other hand, Southern European countries have a lower incidence, respectively: 0.42/100,000 in Spain and 1.07/100,000 in Greece (10).

Observed in own study significant territorial variability of incidence is consistent with those reported in the United States. The highest value (9.45–11.83/100 000 population) concerned north USA regions, while slightly lower was registered in

southern and western US states (7.84–9.43/100,000 population and 4.31–4.93/100,000 population respectively) (21). It is worth referring to the earlier publication, in which authors conclude that observed territorial variability of lung sarcoidosis is related to potentially higher exposure to pesticides and wood dust of people living in districts with a predominance of arable and/or forest land (11). The observations of Italians are also interesting, they pointed to the possible connection between the place of residence in the peripheral areas of cities and rural areas and the increased incidence of lung sarcoidosis (24). However, such hypothesis requires future research in which we have to control exposure.

The average annual cost of sarcoidosis treatment in the Silesian Voivodeship was PLN 2,337.45 (EUR 538.21) per patient. It should be noted a significant disproportion of costs in particular clinical forms of the disease. The lowest cost, on average PLN 775.59 (EUR 178.58) per patient was related to sarcoidosis of skin treatment, while the highest (on average PLN 2,999.80, EUR 688) are related to the lymph nodes sarcoidosis treatment. The cost of hospitalization of sarcoidosis of lung was one of the highest (on average PLN 5,235.45, EUR 1,205.49), and it is worth to notice that in Poland hospitalization of granulomas, allergic and autoimmune pulmonary disease is related with the highest cost of hospitalization in comparison to the most frequent diseases (25). Additionally, the cost of hospitalization was much higher than the cost of outpatient treatment, which is basically associated with the need to finance the simultaneous treatment of co-morbidities and specialist treatment (16). Really, 1/3 of patients with pulmonary sarcoidosis visit a first-time medical doctor with a significant delay (usually half-yearly) in relation to early respiratory symptoms, which causes an increasing cost of treating patients with advanced disease (26). In the end, we should refer to existing problems of health services financing in Poland which are a consequence of insufficient access to guaranteed services. According to the Watch Health Care (WHC) report, the average waiting time in 2017 was dependent on the medical specialization (eg. dermatology needs 2.2 months, internal medicine about 3.1 months, cardiology 5.0 months, and ophthalmology 5.6 months) (27). This situation usually leads to transferring significant costs of diagnosis and treatment to the house budget. It happens that patients ignore symptoms, which ultimately results

in the prolonged diagnostic process and increases the cost of treatment. The matter seems to be important to public health because the last EHCI rankings point to the need for expected reforms in Poland in a range of improvement in the availability and quality of health services provided (28).

CONCLUSIONS

Obtained results confirmed, that sarcoidosis in the Silesian Voivodeship is rather a rare disease with pulmonary sarcoidosis as a dominant clinical form of the disease. The highest number of patients was registered in adults aged 35-54 years, the frequency is higher in men than woman. Significantly decrease of the standardized incidence of sarcoidosis noticed between 2011 and 2015 is related to the observed lower number of total cases of pulmonary disease, and the picture is different than the previous observation registered in years 2006-2010. The average annual unit cost of sarcoidosis hospitalization was significantly higher than the cost of outpatient treatment.

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