

Medical costs of asbestos-related diseases in Spain between 2004 and 2011

Montserrat GARCÍA-GÓMEZ¹, Rosa URBANOS GARRIDO²,
Rosario CASTAÑEDA LÓPEZ¹ and Alfredo MENÉNDEZ-NAVARRO^{3*}

¹Ministry of Health, Social Services and Equality, Spain

²Department of Applied Economics VI. Complutense University of Madrid, Spain

³Department of History of Science, University of Granada, Spain

Received February 13, 2016 and accepted June 14, 2016

Published online in J-STAGE June 23, 2016

Abstract: The objective of this article was to estimate the medical costs derived from malignant ARD treatment in the Spanish National Health System (NHS) between 2004 and 2011. Estimation of direct healthcare costs was based on national primary data on the cost of specialized care for inpatients and outpatients treated at NHS hospitals and on national and regional secondary data on costs of primary healthcare and pharmaceutical prescriptions. A prevalence approach was used to estimate the overall burden of ARDs. Direct medical costs of 37,557 ARDs attended in Spanish NHS facilities in 2004–2011 were estimated at 464 million euros; specialist care accounted for 50.9% of total costs, primary healthcare 10.15%, and drug prescription 38.9%. The cost was 27.8-fold higher in males than in females. Bronchopulmonary cancers represented the greatest healthcare cost, 281 million euros. The cost of delivering healthcare to ARDs victims in Spain has a negative economic impact on the NHS due to the gross under-recognition of occupational victims under the Spanish National Insurance System.

Key words: Asbestos costing, Healthcare economic costs, Asbestos-related diseases, Mesothelioma, Asbestosis, Asbestos-related cancers, Spain

Introduction

Exposure to asbestos fibers increases the risk of disease and death for different causes, but its impact on health is not properly reflected in statistics on occupational disease and must be quantified in specific studies. There have been some local studies of this type¹⁾ and in relation to specific diseases in Spain^{2,3)}, but none in which all asbestos-related diseases (ARDs) were considered.

Exposure to asbestos has specific pathological effects, including asbestosis, the formation of pleural plaques on the parietal pleura with or without calcification, visceral

pleura thickening; benign pleural effusion, and malignant mesothelioma (involving pleura, peritoneum, pericardium, and tunica vaginalis of the testes)⁴⁾. Exposure can also increase the risk of other cancers, notably lung cancer⁵⁾. In 2012, the International Agency for Research on Cancer added the larynx and ovaries as two further cancer sites related to asbestos exposure⁶⁾. In addition, positive associations have been observed between exposure to all forms of asbestos and cancer of the pharynx, stomach, and colorectum⁶⁾.

The WHO has estimated that more than 107,000 people die annually from asbestos-related mesothelioma, lung cancer and asbestosis⁷⁾. In Spain, 2,514,346 tons of asbestos were imported between 1906 and 2001, the year in which the utilization, production, and sale of asbestos fibers and associated products were prohibited⁸⁾. A large

*To whom correspondence should be addressed.

E-mail: amenende@ugr.es

©2017 National Institute of Occupational Safety and Health

Table 1. Matrix of diseases, ICD codes, and attributable fractions due to occupational asbestos exposure by sex

DISEASE	CIE-9	CIE-10	Attributable fraction (%) (95% Confidence interval)			SOURCE
			Males	Females	Total	
Asbestosis	501	J61	100	100	100	WHO ¹²⁾
Mesothelioma	158, 163	C45	97 (96.0–98.0)	82.5 (75.0–90.0)	94.9 (93.0–96.9)	Rushton <i>et al.</i> ¹³⁾
Lung	162	C33-C34	14	0.6	—	Nurminen, Karjalainen ¹⁴⁾
Laryngeal	161	C32	8.3	0.3	—	Nurminen, Karjalainen ¹⁴⁾
Ovarian	183.0	C56	—	0.2–0.5	0.3	Driscoll ¹⁵⁾
Colon	153	C18	2.3	0	—	Nurminen, Karjalainen ¹⁴⁾
Rectum	154	C20	2.9	0	—	Nurminen, Karjalainen ¹⁴⁾

proportion of the imported asbestos was used in buildings, pipes, and roofs constructed between the 1960s and 1990s; hence, the mortality and morbidity burden due to asbestos exposure will continue to rise over the next few years, given the usually long latency period before the appearance of these diseases (e.g., up to 40–50 years for mesothelioma)⁹⁾.

An account was recently published of the asbestos-related occupational diseases recognized by the Spanish National Insurance System between 1961 (when asbestosis was first acknowledged as an occupational disease) and 2010, recording 815 cases of asbestosis¹⁰⁾. In addition, 164 asbestos-related occupational cancers were recorded between 1978, when asbestos-related bronchopulmonary cancers and mesothelioma were first recognized as occupational diseases, and 2011¹¹⁾. These data contrast with the more conservative estimates published in other studies, suggesting that an accurate picture of the true impact of asbestos on health requires the integration of information on patients with asbestosis, pleural plaques, or mesotheliomas treated in the Spanish National Health Service (NHS) and on the population-attributable fraction of tumors known to be associated with asbestos exposure, i.e., in lung, larynx, ovary, colon, and rectum⁶⁾.

The medical costs of treating ARDs have received little attention. The objective of this study was to estimate the burden of malignant ARDs to the Spanish NHS between 2004 and 2011, calculating the direct medical costs of its treatment. Estimation of the burden of a disease and its costs is essential for taking optimal public health decisions in relation to risk reduction/control measures and the planning of human and material healthcare resources.

Methods

The burden (number) of ARDs in Spain between 2004 and 2011 was estimated by identifying all cases due to

asbestosis, almost all cases due to mesothelioma, and the proportion (and therefore number) of cases of lung, laryngeal, ovarian, colon and rectum cancers attributable to asbestos exposure. Table 1 provides a matrix of disease categories [with International Classification of Diseases-9th Revision (ICD-9) codes] and the population-attributable fraction by sex^{12–15)}. These data were combined with healthcare and associated costs obtained from the different NHS care levels. A prevalence approach was used to estimate the overall burden of ARDs. The study period begins in 2004, the first year with available data on specialist outpatient care, and ends in 2011, the latest year for which data are available on each disease type.

Mesotheliomas as a proportion of recorded pleural and peritoneal cancers

The Spanish NHS uses the ICD-9 classification system, which has no specific code for mesothelioma. Therefore, we applied a multiplier obtained in a previous study on the proportion of pleural and peritoneal cancers estimated to be mesotheliomas in Spain between 2004 and 2006⁹⁾, in which 362 out of 496 deaths coded as cancer of the pleura were pleural mesotheliomas, and 51 out of 673 coded as cancer of the peritoneum were peritoneal mesotheliomas. Hence, the number of cases due to pleural mesothelioma was estimated by multiplying the number of pleural cancer cases by 0.68 (0.73 in men and 0.55 in women), and the number due to peritoneal mesothelioma was estimated by multiplying the number of peritoneal cancer cases by 0.08 (0.1 in men and 0.05 in women).

Calculation of the costs

We estimated direct healthcare costs of ARD's for the Spanish NHS. Two types of analysis were performed: a) using NHS data on the cost of specialist (hospital and outpatient) care to compute the cost of cases treated at NHS hospitals; and b) mining secondary data sources to estimate

Table 2. Distribution of number of patients and public healthcare budget across disease categories and types of service in Baix Empordà (euros) and cost factors (relative weights) for primary healthcare and pharmaceutical care

	LUNG		LARYNX		OVARY		COLON/RECTUM	
	n	Mean cost	n	Mean cost	n	Mean cost	n	Mean cost
Primary Care	349	345.8	461	345.8	87	322.3	495	259.9
Pharmaceutical Care	349	1,302.6	461	1,302.6	87	710.5	495	1,418.5
Specialized Care	349	1,652.2	461	1,652.2	87	1,777.6	495	1,826.7
Cost factor for PC (PC/SC)		0.21		0.22		0.18		0.14
Cost factor for PhC (PhC/SC)		0.79		0.60		0.40		0.78

PC=primary care, SC=specialist care, PhC=pharmaceutical care

Source: prepared by the authors from primary data provided by Serveis de Salut Integrats Baix Empordà.

the costs of primary healthcare and prescription drugs^a.

Data in the Information System of the NHS was accessed using the NHS Interactive Consultation program, selecting the registry of hospital discharges (CMBD-H) and registry of specialist outpatient care (CMBD-AAE)¹⁶. The study variables and inclusion criteria were as follows:

- **Years:** from 2004 through 2011;
- **Age and sex:** males or females aged 25 years or older for asbestosis, 40 years or older for mesotheliomas, and 25 years or older for other asbestos-related cancers;
- **Diagnosis:** main diagnosis coded using the ICD-9 system (5 digits);
- **Subjects in the CMBD-H database:** patients discharged from a Spanish public hospital, excluding those with discharge date unavailable or outside the study period, those with admission and discharge on the same day (non-hospitalized), and discarding duplicate records;
- **Subjects in the CMBD-AAE database:** patients receiving specialist outpatient care (sessions, visits, contacts) in day hospital, outpatient department, or at home, excluding those with discharge date unavailable or outside the study period and discarding duplicate records;
- **Mean length of hospital stay;**
- **Cost:** mean estimated cost of treating these patients in a representative sample of NHS hospitals for each year studied;
- **Weighted Activity Unit (WAU):** the unit cost of specialist outpatient care in the years under study, based on the official measure WAU (known in Spain as the *unidad ponderada asistencial UPA*)^{17, 18}.

Estimations of primary healthcare and pharmaceutical

costs were based on data from a Catalonian health district (Baix Empordà) for 2004, 2005, 2006, 2007 and 2011, the years for which cost data were available by clinical risk group. We calculated the costs for primary care and pharmaceutical care as a proportion of costs for specialist care. Primary care included the total use of primary healthcare resources (family doctor, nursing staff or emergencies), pharmaceutical care included all expenditure on prescription drugs, and specialist care included inpatient care, specialist outpatient care and also the hospital pharmaceutical costs, which are included in the CMBD and WAU databases. Information on costs related to long-stay hospitals was not considered because these data are not included in the CMBD database.

Because the Baix Empordà district covers a relatively small population (around 134,000 in 2013), only a small number of patients were recorded in each diagnostic group under study. It would therefore have been inappropriate to calculate the mean cost of each disease per year, which may be heavily influenced by the severity of a given patient. For this reason, we computed the mean cost of each disease over the entire study period and for the whole population (males and females). The ratios obtained were applied to the time-series data on specialist care costs in order to obtain the primary care and pharmaceutical care expenditure.

Table 2 exhibits these calculations, in which the ‘*cost factor for primary care*’ is the primary care expenditure divided by the specialist care expenditure, and the ‘*cost factor for pharmaceutical care*’ is the pharmaceutical care expenditure divided by the specialist care expenditure.

The cost factors for primary care and pharmaceutical care were not available for asbestosis or mesothelioma, because there were no records of patients with these dis-

^aWe are very grateful to Jose M. Inoriza and Marc Carreras from Serveis de Salut Integrats Baix Empordà, who provided us the data, for their valuable assistance.

Table 3. Hospital cost per asbestos-related disease by sex. Spain 2004–2011

Asbestos-related disease (ARD)	Number of hospital discharges		Mean length of stay (days)		Cost per process (in euros)		Cases attributable to occupational exposure		Hospital cost per ARD (in euros)	
	M	F	M	F	M	F	M	F	M	F
Asbestosis										
2004	36	3	11.92	5.33	4,093.89	3,434.31	36	3	147,379.94	10,302.92
2011	28	3	8.61	12	4,128.48	6,344.86	28	3	115,597.45	19,034.57
2004–2011	268	21	9.87	7.48	4,110.27	4,290.10	268	21	1,101,552.15	90,091.99
Mesothelioma										
2004	361	107	14.24	13.97	5,832.20	5,907.79	350	88	2,041,270.00	519,885.52
2011	463	123	11.25	12.13	8,093.18	8,481.00	449	102	3,633,837.82	865,062.00
2004–2011	3,165	960	12.96	13.94	6,815.73	7,228.29	3,07	791	20,924,690.93	5,719,074.03
Malignant neoplasm of trachea, bronchus and lung										
2004	23,041	3,060	11.78	12.40	5,092.20	5,072.77	3,226	18	16,427,451.14	91,309.94
2011	22,868	4,776	9.92	10.13	5,621.33	5,405.07	3,202	29	17,999,483.46	156,747.17
2004–2011	182,476	30,527	11.13	11.39	5,411.89	5,436.92	25,547	183	138,257,940.44	995,173.44
Malignant neoplasm of larynx										
2004	5,152	263	15.07	14.20	7,056.81	6,753.14	428	1	3,020,313.5	6,753.14
2011	4,742	323	12.84	12.76	8,704.33	8,864.76	394	1	3,429,504.72	8,864.76
2004–2011	39,936	2,234	14.46	13.56	8,329.03	8,318.36	3,314	8	27,601,055.38	66,140.42
Malignant neoplasm of ovary										
2004	—	3,704	—	13.22	—	5,506.38	—	11	—	60,570.14
2011	—	4,046	—	10.74	—	8,077.14	—	12	—	96,925.73
2004–2011	—	31,191	—	12.12	—	6,313.99	—	93	—	586,988.37
Malignant neoplasm of colon										
2004	10,765	7,843	16.79	16.68	7,614.87	7,209.04	248	0	1,888,488.06	0
2011	14,519	10,029	13.75	13.06	10,932.31	10,125.43	334	0	3,651,392.64	0
2004–2011	100,491	72,381	15.37	14.92	9,490.83	8,840.00	2,311	0	21,933,092.35	0
Malignant neoplasm of rectum										
2004	7,715	4,504	16.51	16.55	6,831.67	6,594.42	224	0	1,530,293.27	0
2011	8,868	4,949	13.73	13.17	10,030.18	9,441.17	257	0	2,577,757.24	0
2004–2011	67,270	38,205	15.54	14.93	8,651.76	8,158.30	1,951	0	16,877,571.21	0
Total ARD										
2004	47,070	15,780	14.08	18.85	6,174.19	7,894.71	4,512	121	25,055,195.91	688,821.66
2011	51,488	20,203	11.94	14.53	8,183.68	10,428.77	4,664	147	31,407,573.33	1,146,634.23
2004–2011	393,606	144,328	13.32	16.77	7,313.38	9,284.83	36,461	1,096	226,695,902.46	7,457,468.25

Source: prepared by the authors from data provided by the National Health System “Consulta Interactiva”, Ministerio de Sanidad, Servicios Sociales e Igualdad. Instituto de Información Sanitaria (2013)¹⁶⁾

eases in Baix Empordà during the analyzed period. Therefore, the cost factor obtained for lung cancer was used for these diseases, on the grounds that their medical costs would be relatively similar.

Results

Between 2004 and 2011, there were 37,557 admissions for ARDs to Spanish public hospitals of individuals over the age of 25 years (>40 years in the case of mesotheli-

oma), 97.1% of whom were male and 2.9% female. The rate of hospital discharges for ARDs per 100,000 Social Security-affiliated employees was 43.9 in males and 1.9 in females. Over this period, men showed a slight increase in this rate (43.9 in 2004 vs. 49.2 in 2011), while women evidenced stability (1.8 in 2004 vs. 1.9 in 2011). Hospital admissions for mesothelioma increased, especially among males (350 occupational-related cases in 2004 vs. 449 in 2011), while admissions for asbestosis decreased (39 in 2004 vs. 31 in 2011) (Table 3).

Table 4. Cost of specialized outpatient care per asbestos-related disease by sex. Spain 2004–2011

Asbestos-related disease (ARD)	Number of consultations in specialist outpatient care		Weighted activity unit cost (in euros)		Specialist outpatient care consultations attributable to occupational exposure		Specialist outpatient care cost per ARD (in euros)	
	M	F	M	F	M	F	M	F
Asbestosis								
2004	0	0	0	0	0	0	0	0
2011	2	0	359.74	0	2	0	719.49	0
2004–2011	15	4	308.61	481.36	15	4	4,629.20	1,925.43
Mesothelioma								
2004	5	2	295.85	299.52	5	1	1,479.23	299.52
2011	62	35	472.20	416.07	60	28	28,331.96	11,649.87
2004–2011	196	93	396.28	369.59	191	75	75,595.26	27,703.06
Malignant neoplasm of trachea, bronchus and lung								
2004	852	190	324.25	306.87	119	1	38,585.41	306.87
2011	4,507	1,399	424.91	400.20	631	8	268,117.36	3,201.62
2004–2011	22,765	5,438	379.94	373.61	3,188	33	1,211,236.48	12,325.12
Malignant neoplasm of larynx								
2004	363	23	351.23	356.74	30	0	10,536.81	0
2011	1,175	68	508.62	520.86	98	0	49,844.41	0
2004–2011	5,990	370	454.05	494.74	498	0	226,139.29	0
Malignant neoplasm of ovary								
2004	—	393	—	312.45	—	1	—	312.45
2011	—	1,516	—	564.20	—	5	—	2,821.01
2004–2011	—	7,290	—	426.55	—	22	—	9,548.98
Malignant neoplasm of colon								
2004	545	389	340.18	324.10	13	0	4,422.40	0
2011	6,046	3,697	596.40	552.38	139	0	47,285.68	0
2004–2011	30,599	19,699	499.10	464.88	705	0	239,830.24	0
Malignant neoplasm of rectum								
2004	289	202	310.37	298.78	8	0	2,482.95	0
2011	3,433	1,865	548.04	515.86	100	0	54,804.48	0
2004–2011	17,947	10,121	453.21	449.67	522	0	236,610.88	0
Total ARD								
2004	2,054	1,199	331.22	313.87	175	3	57,506.80	918.84
2011	15,225	8,580	527.42	375.36	1,030	41	449,103.38	17,672.5
2004–2011	77,512	43,015	449.70	344.78	5,119	134	1,994,041.35	51,502.59

Source: prepared by the authors from data provided by the National Health System “Consulta Interactiva”, Ministerio de Sanidad, Servicios Sociales e Igualdad. Instituto de Información Sanitaria (2013)¹⁶

Between 2004 and 2011 there were 5,253 specialist outpatient visits for the diseases under study, 97.4% by men and 2.6% by women (Table 4). Although the male-female ratio differed among the diseases, there was always a predominance of males. Cases attended in day hospitals were 6-fold higher in 2011 than in 2004, with a 14-fold increase for females, and an increase was observed in both sexes for all diseases under study with the exception of asbestosis.

Hospital cost per disease was highest for colon and rectum cancers, which also required a longer hospital

stay. The mean length of hospital stay tended to decrease over the study period for all diseases in both sexes except for asbestosis in women. The lowest hospital costs were for asbestosis. The cost was higher in women for mesothelioma (Table 3). The total public hospital cost for the eight-year study period was 234,153,370.71 €, of which 96.8% corresponded to male patients. Between 2004 and 2011, these costs rose by 25.3% for men and by 66.5% for women. The total cost of specialist outpatient care for these diseases was 2,045,543.95 € over the same period

Table 5. Total healthcare cost per asbestos-related disease by type of service and sex (euros). Spain 2004–2011

Asbestos-related disease (ARD)	Specialist care cost		Primary care cost		Pharmaceutical care cost		Healthcare cost per ARD	
	M	F	M	F	M	F	M	F
Asbestosis								
2004	147,379.94	10,302.92	30,949.79	2,163.61	116,430.15	8,139.31	294,759.88	20,605.84
2011	116,316.93	19,034.57	24,426.56	3,997.26	91,890.37	15,037.31	232,633.86	38,069.14
2004–2011	1,106,181.35	92,017.41	232,298.08	19,323.66	873,883.27	72,693.75	2,212,362.70	184,034.82
Mesothelioma								
2004	2,042,748.22	520,184.67	428,977.13	109,238.78	1,613,771.09	410,945.89	4,085,496.44	1,040,369.34
2011	3,662,168.71	876,712.17	769,055.43	184,109.56	2,893,113.28	692,602.61	7,324,337.42	1,753,424.34
2004–2011	21,000,285.21	5,746,776.18	4,410,059.89	1,206,822.00	16,590,225.32	4,539,953.18	42,000,570.42	11,493,552.36
Malignant neoplasm of trachea bronchus, and lung								
2004	16,466,036.55	91,616.81	3,457,867.68	19,239.53	13,008,168.87	72,377.28	32,932,073.10	183,233.62
2011	18,267,600.82	159,948.79	3,836,196.17	33,589.25	14,431,404.65	126,359.54	36,535,201.64	319,897.58
2004–2011	139,469,176.91	1,007,498.57	29,288,527.15	211,574.70	110,180,649.76	795,923.87	278,938,353.82	2,014,997.14
Malignant neoplasm of larynx								
2004	3,030,850.31	6,753.14	666,787.07	1,485.69	1,818,510.19	4,051.88	5,516,147.56	12,290.71
2011	3,479,349.13	8,864.76	765,456.81	1,950.25	2,087,609.48	5,318.86	6,332,415.42	16,133.86
2004–2011	27,827,194.66	66,140.42	6,121,982.83	14,550.89	16,696,316.80	39,684.25	50,645,494.28	120,375.56
Malignant neoplasm of ovary								
2004	—	60,882.59	—	10,958.87	—	24,353.04	—	96,194.49
2011	—	99,746.74	—	17,954.41	—	39,898.70	—	157,599.85
2004–2011	—	596,537.34	—	107,376.72	—	238,614.94	—	942,528.00
Malignant neoplasm of colon								
2004	1,892,910.46	0	265,007.46	0	1,476,470.16	0	3,634,388.08	0
2011	3,698,678.31	0	517,814.96	0	2,884,969.08	0	7,101,462.36	0
2004–2011	22,172,922.58	0	3,104,209.16	0	17,294,879.61	0	42,572,011.35	0
Malignant neoplasm of rectum								
2004	1,532,776.22	0	214,588.67	0	1,195,565.45	0	2,942,930.34	0
2011	2,632,561.72	0	368,558.64	0	2,053,398.14	0	5,054,518.50	0
2004–2011	17,114,182.11	0	2,395,985.50	0	13,349,062.05	0	32,859,229.65	0
Total ARD								
2004	25,112,701.70	689,740.13	5,064,177.79	143,086.48	19,228,915.92	519,867.40	49,405,795.41	1,352,694.01
2011	31,856,675.62	1,164,307.03	6,281,508.57	241,600.72	24,442,385.00	879,217.02	62,580,569.19	2,285,124.77
2004–2011	228,689,942.82	7,508,969.92	45,553,062.61	1,559,648.97	174,985,016.80	5,686,869.99	449,228,022.23	14,755,488.88

Source: prepared by the authors from Tables 1–4.

(Table 4) and evidenced the greatest rise in costs between 2004 and 2011, with a 19.2-fold increase in females and 7.8-fold increase in males. Globally, the total cost of specialist healthcare (hospital + outpatient) was 236,198,914.7 €.

Table 5 displays the annual costs of healthcare services and prescriptions for the seven ARDs studied between 2004 and 2011. The total cost over the 8-year period was 463,983,511 €, being 30.4-fold higher for male vs. female cases. This total cost began to rise after 2006, increasing by 27.8% between 2004 and 2011 (26.7% for males vs. 68.9% for females). This increase was not uniform over the period and even decreased slightly in 2005 and 2010. Among the

years showing an inter-annual increase, this ranged from 2.5% between 2008 and 2009 to 8.9% between 2006 and 2007. The only ARD evidencing a reduction in total costs over the study period was asbestosis in men. These inter-annual changes were relatively similar among the three types of healthcare considered. The highest costs incurred over the period were for tracheal and bronchopulmonary cancers, both globally (60.5%) and among the men (62.1%), whereas mesothelioma incurred the highest costs among the women (77.9% of total for females). Specialist healthcare represented 50.9% of the total healthcare cost of these ARDs. The next highest cost was that for pharmaceutical care, which represented 38.95% of the total, while

Table 6. Mean age (SD) and mean healthcare cost per asbestos-related disease by sex (euros). Spain 2004–2011

Asbestos-related disease (ARD)	Mean age (SD)		Mean healthcare cost per ARD (in euros)	
	M	F	M	F
Asbestosis				
2004	72.8 (7.4)	80.3 (2.9)	8,187.77	6,868.61
2011	73 (10.2)	73.6 (10.4)	8,308.35	12,689.71
2004–2011	72.6 (8.9)	74.3 (13.1)	8,307.78	9,057.85
Mesothelioma				
2004	67.5 (10.7)	67.2 (13.2)	11,672.85	11,822.38
2011	69.4 (10.6)	69.1 (11.6)	16,312.56	17,190.43
2004–2011	68.6 (10.3)	68.9 (12.1)	13,571.92	14,444.55
Malignant neoplasm of trachea, bronchus, and lung				
2004	66.2 (11)	62.5 (13.8)	10,208.33	10,179.65
2011	67.5 (10.7)	64 (12.9)	11,410.12	11,030.95
2004–2011	66.8 (10.9)	63.2 (13.3)	10,914.89	10,935.89
Malignant neoplasm of larynx				
2004	62.7 (10.8)	57.4 (13.5)	12,888.20	12,290.71
2011	64.1 (10.6)	61.5 (12.6)	16,072.12	16,133.86
2004–2011	63.4 (10.8)	60 (12.8)	15,294.45	15,046.94
Malignant neoplasm of ovary				
2004	—	61.5 (14.1)	—	8,744.95
2011	—	61.3 (14.3)	—	13,133.32
2004–2011	—	61.1 (14.3)	—	10,086.77
Malignant neoplasm of colon				
2004	69.5 (11.1)	—	14,654.79	0.00
2011	70.4 (11.2)	—	21,261.86	0.00
2004–2011	70.1 (11.2)	—	18,199.75	0.00
Malignant neoplasm of rectum				
2004	68.5 (11.2)	—	13,138.08	0.00
2011	68.6 (11.6)	—	19,667.39	0.00
2004–2011	68.6 (11.4)	—	16,720.29	0.00

Source: prepared by the authors from data provided by Ministerio de Sanidad, Servicios Sociales e Igualdad (2013)¹⁶ and Table 5

primary healthcare was responsible for 10.15% of the total costs.

Finally, Table 6 lists the mean costs per case for each ARD in 2004 and 2011 and in the whole period under study (2004–2011) as well as the mean age (SD) of hospital inpatients, the only patients for whom these data are available.

Discussion and Conclusion

The historic use and handling of asbestos in Spain continue to evidence a major impact on health. ARDs produce around 4,700 hospital admissions and over 650 specialist outpatient consultancies per year, and their direct annual cost to the Spanish NHS is estimated to be around 58 mil-

lion euros. In fact, this is an underestimation of the global expenditure by the NHS on ARDs, given that the fraction of pharynx and stomach cancers attributable to asbestos has not been considered. In industrialized countries, occupational exposure is responsible for 100% of asbestosis cases, at least 85% of mesothelioma cases, and a variable proportion of the other diseases studied. Between 2004 and 2011, 37,557 public hospital admissions were for ARDs in individuals aged 25 years or more (>40 yrs for cases of mesothelioma), mainly for lung cancer, mesothelioma, or laryngeal cancer. Furthermore, 2,611 individuals aged 40 years or older (1,952 men and 659 women) died from pleural mesothelioma over the eight-year study period¹⁶. In contrast, only 490 cases of ARDs were recognized as occupational diseases under the Spanish National Insurance System over the same period: 81 cases of mesotheliomas, 66 of bronchopulmonary cancer, 280 of asbestosis, and 63 of pleural plaques^{10,11}. Evaluating the costs of ARDs based on workers' compensation can lead to gross underestimations and hinder comparison between countries, because of differences in national legislative frameworks, insurance systems, and recognition rates. Thus, compensation schemes cover 35% of mesothelioma cases in Italy and between 16% and 25% of those in France, depending on the region¹⁹, whereas only 6.4% of mesothelioma cases in men and 0.3% of those in women are considered as occupational diseases in Spain¹¹. The gross under-recognition of ARDs in Spain not only reveals the deficiencies of the Spanish National Insurance System but also has a negative economic impact on the NHS, which is estimated to incur healthcare costs of 58 million euros every year that should be paid under the compensation scheme for occupational ARDs victims. It is estimated that over a thousand individuals will die from pleural mesothelioma in Spain between 2016 and 2020, and asbestos-related deaths are expected to continue until at least 2040⁹. For this reason, the gathering, analysis, and diffusion of data on these diseases and on their relationship with occupational exposure is crucial for planning prevention and healthcare programs and for the appropriate compensation of victims. The calculations and analysis of the healthcare burden of ARDs in our study represent useful tools for informing decisions and redefining health policies and the human and material resources required.

More than half (68.5%) of hospital admissions for ARDs were for bronchopulmonary cancers, followed by mesothelioma (10.3%) and laryngeal cancer (8.8%), while the smallest number was for asbestosis (0.8%), similar to previous findings in Europe²⁰. We highlight the marked

increase in specialist outpatient consultancies for ARDs from 178 in 2004 to 1,071 in 2011, which may be attributable to a trend for the outpatient rather than in-hospital treatment of these diseases. It should be borne in mind that the coverage of the information system is complete for hospitalizations and for specialist outpatient surgery but is only 46.7% for the medical activity of the outpatient day hospital²¹). A further potential limitation is that our utilization of ratios for the whole period as a proxy of primary, specialist, and pharmaceutical care costs assumes that there was no substantial variation in the treatment of these diseases between 2004 and 2011. This appears to be a reasonable assumption, but a certain degree of bias cannot be ruled out.

There was an increase in consultancies for all ARDs with the exception of asbestosis, responsible for the smallest number of consultancies, which rose until 2006 and then declined, consistent with published evidence on the decline in the morbidity and mortality of this disease in Spain and Europe^{10,20}).

Previous studies on the attribution of ARDs to occupational exposure concluded that this approach is highly unlikely to overestimate the true impact of this exposure on morbidity and mortality^{22–24}). In fact, as noted above, our data likely represent an underestimation, because no account was taken of pharyngeal or stomach cancers, as the proportion due to asbestos exposure has not yet been elucidated. The WHO concluded that occupational exposure to asbestos remains a major cause of death and disability in Europe, despite the conservative assumptions that underlie published estimates¹²). All studies have found that more than half of occupational cancers involve the lung, and that asbestos is the main occupational carcinogen²⁵).

We calculated the direct costs to the NHS of the diagnosis and treatment of the disease but took no account of indirect costs, such as the loss of productivity caused by the temporary/permanent disability and premature death of workers or the cost of carers, etc. Even though our estimate of the total costs generated by these conditions is therefore at the lower end, it indicates an expenditure of more than 463 million euros between 2004 and 2011 (51 in 2004 and 65 in 2011), with a mean annual increase of 3.5%. It has been reported that 66%–85% of the total health costs of cancer are indirect costs, including sick leave and early retirement, and especially the cost of mortality in people of working age^{12,26–28}). Evaluation of the full economic impact of ARDs in Spain requires an assessment of the indirect costs and their addition to the direct costs calculated in the present study.

The greatest proportion of direct healthcare costs were for specialist care services (around 51%), followed by pharmaceutical prescriptions (39%). It was previously reported that 50% of the cost of cancer in Spain was for the hospitalization of patients, and that 15% was for pharmacologic treatments. Over the period from 2004 through 2011, there was no difference in the time course of costs among the components of specialist care with the exception of day hospital costs, which showed a mean annual increase of more than 100% (10-fold higher in 2011 than in 2004), whereas the average annual increase in hospitalization was only 2.9%.

Comparison of our findings is hampered by the different methods used in research on medical costs as a function of the specific objectives and resources available, as affirmed at the WHO meeting in Bonn in 2012¹²). The annual global cost of medical treatment for ARDs has been estimated at 4.34 billion USD²⁹). At the aforementioned WHO meeting in Bonn, it was stated that the mean acute treatment cost per case of mesothelioma was 15,899.34 € in the United Kingdom¹²), compared with previous estimates of 15,000 € in Scotland in 2006³⁰), 24,000 € in Italy in 2011¹⁹), and 20,218 € for males in 2011 (present study). Lifetime healthcare costs of pleural mesothelioma in Taiwan were estimated to be 14,900 USD in 1997–2005³¹). An investigation of the direct healthcare sources of occupational lung and bladder cancers in Spain in 2008²⁴) estimated a cost per lung cancer patient of 9,277 €, lower than the cost of 11,566 € estimated in the present study. In Canada, the median cost per case in the 2000s of non-small cell lung cancer and small cell lung cancer were \$10,928 (range \$9,234–11,047) and \$15,350 (range \$13,033–21,436) respectively¹²). The aforementioned study on cancer costs in Spain found a cost of 19,535 € per case of colon cancer²⁷), similar to the present finding although different computational methods were used.

Our calculation of the direct healthcare costs of occupational ARDs in Spain should be considered the lower limit for the reasons noted above, but it is not possible to calculate the degree of underestimation from the information available. The assumptions used to calculate the components of healthcare also represent a bias, especially with respect to the costs of prescriptions and primary care, whose estimation was derived from a geographic area that cannot be considered representative of the nation as a whole, given the variability in disease prevalence and in the offer of medical services among regions. Nevertheless, there can be no doubt about the high healthcare costs of ARDs in Spain, especially given the potentially prevent-

able nature of these diseases. The elevated economic burden of ARDs underlines the need to increase their prevention and their early diagnosis and treatment.

As declared by the WHO European Region at its latest meeting¹²⁾, asbestos continues to present a public health threat, despite bans on its import and use, due to wide exposure in the past. The recognition, diagnosis, and recording of ARDs remain challenging. However, estimation of their costs is of particular importance at a time of economic pressures on the health service. The proper demarcation of financial responsibilities between the National Insurance System (including Work Accidents and Occupational Diseases Insurance Institutes) and the National Health System is a highly relevant issue. Costs of occupational diseases should be funded by social contributions from employers and employees under the National Insurance System rather than by the tax-funded National Health System.

Acknowledgements

This study was funded by the Spanish Ministry of Science and Innovation (Project HAR2009-07543).

References

- 1) Tarrés J, Abós-Herrándiz R, Albertí C, Martínez-Artés X, Rosell-Murphy M, García-Allas I, Krier I, Castro E, Cantarell G, Gallego M, Orriols R (2009) Enfermedad por amianto en una población próxima a una fábrica de fibrocemento. *Arch Bronconeumol* **45**, 429–34.
- 2) Agudo A, González CA, Bleda MJ, Ramírez J, Hernández S, López F, Calleja A, Panadès R, Turuguet D, Escolar A, Beltrán M, González-Moya JE (2000) Occupation and risk of malignant pleural mesothelioma: A case-control study in Spain. *Am J Ind Med* **37**, 159–68.
- 3) Badorrey MI, Monsó E, Teixidó A, Pifarré R, Rosell A, Llatjós M (2001) Frecuencia y riesgo de neoplasia broncopulmonar relacionada con asbestos. *Med Clin (Barc)* **117**, 1–6.
- 4) Ministerio de Sanidad, Servicios Sociales e Igualdad (2013) Protocolos de Vigilancia Sanitaria Específica: Amianto, 3ª ed., 61, Ministerio de Sanidad, Servicios Sociales e Igualdad, Madrid.
- 5) IARC (1987) Overall evaluations of carcinogenicity: an updating of IARC Monographs volumes 1 to 42. *IARC Monogr Eval Carcinog Risks Hum Suppl* **7**, 1–440 [IARC, Lyon.].
- 6) IARC (2012) A Review of Human Carcinogens. Part C: Arsenic, metals, dusts, and fibres. *IARC Monogr Eval Carcinog Risks Hum* **100C**, 501 [IARC, Lyon.].
- 7) World Health Organization (2010) Asbestos: Elimination of Asbestos-Related Diseases, Fact sheet no. 343, WHO, Paris. <http://www.who.int/mediacentre/factsheets/fs343/en/index.html>. Accessed March 17, 2015.
- 8) Calleja i Vila A, Hernández Carrascosa S (2001) Amiant en edificis i instal·lacions Què fer? Generalitat de Catalunya, Departament de Treball, Barcelona [cd].
- 9) López-Abente G, García-Gómez M, Menéndez-Navarro A, Fernández-Navarro P, Ramis R, García-Pérez J, Cervantes M, Ferreras E, Jiménez-Muñoz M, Pastor-Barriuso R (2013) Pleural cancer mortality in Spain: time trends and updating of predictions up to 2020. *BMC Cancer* **13**, 528.
- 10) García Gómez M, Menéndez-Navarro A, Castañeda López R (2012) Incidencia en España de la asbestosis y otras enfermedades pulmonares benignas debidas al amianto durante el período 1962–2010. *Rev Esp Salud Publica* **86**, 613–25.
- 11) García-Gómez M, Menéndez-Navarro A, López Castañeda R (2015) Asbestos-related occupational cancers compensated under the Spanish National Insurance System, 1978–2011. *Int J Occup Environ Health* **21**, 31–9.
- 12) World Health Organization (2013) The human and financial burden of asbestos in the WHO European Region. Report on a meeting held in Bonn, 5–6 November 2012, 85, WHO, Copenhagen.
- 13) Rushton L, Hutchings SJ, Fortunato L, Young C, Evans GS, Brown T, Bevan R, Slack R, Holmes P, Bagga S, Cherrie JW, Van Tongeren M (2012) Occupational cancer burden in Great Britain. *Br J Cancer* **107** Suppl 1, S3–7.
- 14) Nurminen M, Karjalainen A (2001) Epidemiologic estimate of the proportion of fatalities related to occupational factors in Finland. *Scand J Work Environ Health* **27**, 161–213.
- 15) Driscoll T (2012) Personal communication. In: World Health Organization. The human and financial burden of asbestos in the WHO European Region. Report on a meeting held in Bonn, 5–6 November 2012.
- 16) Ministerio de Sanidad, Servicios Sociales e Igualdad. Instituto de Información Sanitaria (2013) Consulta Interactiva del SNS. <http://pestadistico.inteligenciadegestion.msssi.es>. Accessed February 28, 2015.
- 17) Bestard JJ, Sevilla F, Corella MI, Elola Somoza J (1993) La unidad ponderada asistencial (UPA): nueva herramienta para la presupuestación hospitalaria. *Gac Sanit* **39**, 263–73.
- 18) Ministerio de Sanidad, Política Social e Igualdad. Instituto de Información Sanitaria (2010) Estadística de establecimientos sanitarios con régimen de internado. http://www.msssi.gob.es/estadEstudios/estadisticas/docs/SIAE_Resultadosprovisionales_2010.pdf. Accessed May 3, 2015.
- 19) Merler E, Bressan V, Bilato AM, Marinaccio A; Gruppo regionale veneto sui mesoteliomi maligni (2011) The effectiveness of compensation system for mesotheliomas due to occupational exposure to asbestos and determinants for requests and awards: an evaluation based on record-linkage between the Veneto Mesothelioma Register and the claims and compensations recorded by the National Insurance Institute (INAIL). *Epidemiol Prev* **35**, 331–8.
- 20) Eurogip (2006) Les maladies professionnelles liées à

- l'amiante en Europe. Reconnaissance – Chiffres - Dispositifs spécifiques, 47, Eurogip, Paris.
- 21) Ministerio de Sanidad, Política Social e Igualdad. Instituto de Información Sanitaria (2011) Estadística de establecimientos sanitarios con régimen de internado. Evolución 2000–2008. <http://www.mspsi.gob.es/estadEstudios/estadisticas/estHospiInternado/inforAnual/homeESCRI.htm>. Accessed March 12, 2015.
 - 22) Kogevinas M, Castaño-Vinyals G, Rodríguez Suárez MM, Tardón A, Serra C (2008) Estimación de la incidencia de la mortalidad por cáncer laboral en España, 2002. *Arch Prev Riesgos Labor* **11**, 180–7.
 - 23) García García AM, Gadea Merino R, López Martínez V (2007) Estimación de la mortalidad atribuible a enfermedades laborales en España, 2004. *Rev Esp Salud Pública* **81**, 261–70.
 - 24) García Gómez M, Urbanos Garrido R, Castañeda López R, López Menduina P (2012) Costes sanitarios directos de las neoplasias de pulmón y vejiga de origen laboral en España en 2008. *Rev Esp Salud Pública* **86**, 127–38.
 - 25) Straif K (2008) The burden of occupational cancer. *Occup Environ Med* **65**, 787–8.
 - 26) World Health Organization (2004) Priority Medicines for Europe and the World. http://whqlibdoc.who.int/hq/2004/WHO_EDM_PAR_2004.7.pdf. Accessed March 15, 2015
 - 27) Antoñanzas Villar F, Oliva J, Velasco M, Zozaya N, Lorente R, López Bastida J (2006) Costes directos e indirectos del cáncer en España. *Cuadernos Económicos de ICE* **72**, 281–309.
 - 28) Serrier H, Sultan-Taieb H, Luce D, Bejean S (2014) Estimating the social cost of respiratory cancer cases attributable to occupational exposures in France. *Eur J Health Econ* **15**, 661–73.
 - 29) Aljunid S, Qureshi AM, Baguma D, Kouadio IK. Toolkit for Elimination of Asbestos Related Diseases. Chapter 4: Assessment of Asbestos Production and Consumption with Associated Health and Economic Burden. <http://envepi.med.uoeh-u.ac.jp/toolkit/section4.html>. Accessed June 7, 2016.
 - 30) Watterson A, Gorman T, Malcolm C, Robinson M, Beck M (2006) The economic costs of health service treatments for asbestos-related mesothelioma deaths. *Ann N Y Acad Sci* **1076**, 871–81.
 - 31) Lee LJH, Chang YY, Liou SH, Wang JD (2012) Estimation of benefit of prevention of occupational cancer for comparative risk assessment: methods and examples. *Occup Environ Med* **69**, 582–6.