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ORIGINAL ARTICLE

Reducing the underreporting of lung cancer attributable to occupation: outcomes from a hospital-based systematic search in Northern Italy

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Abstract

Purpose Occupational exposure to lung carcinogens is and was common in workplaces. 5–25 % of lung cancers (LCs) could be causally attributable to occupation; however, LC underreporting and undercompensation are widespread, with remarkable tolls paid by individuals and society. This work aims to: describe an ongoing hospital-based systematic search (SS) of occupational LC; improve aetiological diagnosis; increase number and quality of LC notifications.

Methods Through a short form, physicians at a public hospital referred incident LC to the Occupational Health Unit (OHU). Only patients selected through the form were interviewed; a personal, occupational and clinical history was collected; reports were sent to the ward and Local Health Authority, with aetiological diagnosis criteria and probability of causation.

Results From 1998 to 2013, 3274 cases of LC were notified to the OHU; prior to the system, just couple of dozens were assessed. A total of 1522 patients were fully interviewed; in 395 cases, causation was attributed to occupation (26 % of interviewed patients); all were notified to authorities, as compared to the handful reported before the system was adopted. Main aetiological agents were silica, asbestos, polycyclic aromatic hydrocarbons, truck driving, painting, multiple exposures. Compensation rate was remarkable (39 %).

Conclusions Through SS, many occupational LCs were found that otherwise would have been lost. Aetiological diagnosis proved to be rich of scientific advantages and practical implications, with attention to equity and social aspects. SS was easy, accountable and fostered multidisciplinary collaboration among medical specialties, significantly reducing underreporting and undercompensation of occupational LC.

Keywords Occupational lung cancer · Occupational health physician · Underreporting · Undercompensation · Systematic search

Introduction

Lung cancer (LC) is a leading cause of death worldwide (World Health Organization-WHO and International Agency for Research on Cancer-IARC 2014). In Italy, LC mortality for males is about 23,000 cases/year, with a standardized incidence ratio (SIR) of 59/100,000 (Italian National Institute of Tumours 2015).

Many workers have been or are exposed to occupational lung carcinogens in workplaces (Kauppinen et al. 2000; Scarselli et al. 2007), and dozens of agents, mixtures, exposure circumstances are classified as certain occupational lung carcinogens (International Agency for Research on Cancer-IARC 1979–2015).

Years ago, a milestone paper from Doll and Peto (1981) reported that 2–8 % of all cancer deaths could be caused by occupation: the estimated work-related attributable fraction

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for LC was 15 % for males (M). Several other estimates of occupational LC were published afterwards: 8 % worldwide; 10–33 % (M), USA; 7 % (M), Europe; 29 % (M), Finland; 13–29 % (M), France; 21 % (M), Great Britain; 5–36 %, Italy. Overall, LC accounts for more than half of the occupational related cancer burden and, therefore, very many LCs could be attributable to occupation, thus entitled to be notified to authorities and possibly compensated (De Matteis et al. 2008, 2012; Driscoll et al. 2005; Straif 2008; World Health Organization-WHO 2009). Unfortunately, underreporting and undercompensation of occupational cancers are well known: various scientific contributions analysed these topics, highlighting advantages from proper notification and compensation schemes, as well as need for new intervention schemes to reduce underestimation and to improve current reporting practices and compensation policies (Azaroff et al. 2002; Curti et al. 2015; Leem et al. 2010; Porru et al. 2006; Rosenman et al. 2006; Schulte 2005). Literature data show large discrepancy between what is reported and compensated according to national schemes and what would be expected according to attributable fractions for occupational LC. Overall, the degree of underestimation is of various orders of magnitude and, in Italy, could be more than 80 % (Brugere and Naud 2003; Eurogip 2015; Scarselli et al. 2009; Straif 2008).

Therefore, we may well refer to ‘lost occupational cancers’, for two main reasons:

(a) limited/absent recognition of work-relatedness of LC (i.e. aetiological diagnosis);
(b) limited/absent notification of the work-related LC to health and compensation/insurance authorities (Azaroff et al. 2002; Fan et al. 2006; Friedman and Forst 2007; Porru et al. 2006; Scarselli et al. 2009).

Simple considerations show that if the incidence of LC in Italy is approximately 27,000 new cases/year in males (Italian National Institute of Tumours 2015) and attributable fraction to occupation is 10–15 % (De Matteis et al. 2008, 2012; Doll and Peto1981; Driscoll et al. 2005; Straif 2008; World Health Organization-WHO 2009), the number of expected occupational LC should be 2700–4050; however, only 255–261 LCs were yearly notified, and an occupational causation was recognized for far less (i.e. 184–188, 72 % of LC notified cases). No data are available for compensation rates (Bottazzi 2015; Italian Workers’ Compensation Authority-INAIL 2015).

Limited literature data are available aiming at finding and reporting lost occupational cancer and LC in particular (Ahn and Jeong 2014; Cellier et al. 2013; Crosignani et al. 2006; De Lamberterie et al. 2002; Legrand Cattan et al. 2000; Morelle et al. 2014; Pairon et al. 2006; Slåstad et al. 2014; Spreeuwers et al. 2009). Overall, it is clear that current surveillance and reporting systems miss a large number of occupational LC; therefore, too many workers have no access to social benefits and compensation schemes, and preventive interventions are hampered.

Aims of this work were: (a) to describe a hospital-based systematic search (SS) of occupational LC, by this term meaning an organized, evidence-based system to find, assess, document and report LC, carried out in a highly industrialized territory, with a significant incidence rate of LC; (b) to improve aetiological diagnosis, by increasing number and quality of notifications of occupational LC; (c) to highlight outcomes and advantages of SS from clinical, epidemiological, preventive, individual and public health standpoints; (d) to underline motivations and roles of the occupational physicians (OP) in SS of occupational cancers and LCs in particular.

Methods

In 1998, a still ongoing programme of SS for LC has begun at a public general hospital in Northern Italy, where a significant share of LC occurring in the local population is referred for diagnosis and treatment.

When a primary LC was histologically diagnosed, a single-page form (see supplementary online information) with personal data, diagnosis, information on tobacco smoking and general lifetime occupational history (including main job titles, tasks and occupational sector) was completed by physicians belonging to Pneumology, Thoracic Surgery, Internal Medicine and Radiotherapy wards. The form was sent to the Unit of Occupational Health (OHU) for formal OP evaluation. The OP (always the same) could opt to: file the case, when no occupational exposure to lung carcinogens was apparent (e.g. lifetime administratives, housewives); integrate through direct interview (e.g. to check some temporal variables; to clarify uncertain exposures); proceed for full occupational history evaluation for patients with potential exposure to known or suspected occupational risk factors for LC (e.g. foundry workers, electroplaters, truck drivers, welders, painters, rubber workers, subjects exposed to asbestos, crystalline silica, diesel exhausts, polycyclic aromatic hydrocarbons, certain metals). Histories were taken by direct interview generally from the same OP or residents in occupational medicine, with direct supervision from the OP evaluating the forms. Lifelong smoking habits were fully recorded to divide subjects in non-smokers, former (quit >5 years before diagnosis), current smokers (1 cigarette/day for 1 year at least and those who quit smoking <5 years before diagnosis).

Lifetime occupational history covered every job task and occupational sector, focussing on exposures to all known or suspected occupational lung carcinogens. If needed, further data were acquired on specific work environments or technological cycles, through available industrial hygiene data, safety data sheets, employers, Local Health Authorities.

For each case, pertinent clinical documentation was acquired from each ward or patient. If necessary, imaging were re-evaluated by trained radiologists and the OP, to evaluate interstitial disorders or lesions referable to previous exposure to asbestos or silica. If needed, available histological preparations from pulmonary parenchyma were acquired and re-evaluated by the pathologist to search for asbestos bodies or occupational interstitial disorders.

A detailed clinical report was sent to the hospital ward that diagnosed the LC and to the Local Health Authority, which included criteria for aetiological diagnosis, probability of causation and reference to specific literature supporting the causal attribution. An occupational LC was recognized by the same OP on a case-by-case basis, following criteria for causal attribution as applied in occupational health (Hill 2015; Porru et al. 2006; Verbeek 2012; Wakeford and McElvenny 2007). After aetiological diagnosis, medico-legal obligations were carried out by the physicians of that ward, such as: reporting to Judiciary and notification to Local Health Authority, mandatory for every physician in any operational context; medical certificates for Italian Workers' Compensation Authority (INAIL—which covers almost every formal worker), obligation for every physician to write the certificate and give it to the worker, who then files it to INAIL according to current mandatory Italian laws (Scarselli et al. 2009). Such obligations are free of charge for the worker; the hospital personnel and OP involved in the SS are on staff and not specifically paid or funded for this ongoing programme, which is part of everyday hospital tasks.

After OHU, INAIL and workers' patronage (INCA CGIL) consultations, it was possible to follow up only a few LC cases along compensation and judicial processes. It should be finally noted here that, in Italy, occupational diseases can be notified to INAIL both whether they belong to a national list of occupational diseases that can be compensated and when the causal relationship is certified by physicians, even for disorders not included in the list. For a case to be entitled for compensation, the Italian compensation context requires that occupation can simply be demonstrated as a causal factor in the probability of causation assessment, regardless presence of other causal factors.

The work has been approved by the appropriate institutional review boards and subjects gave informed consent.

Results

The study was carried out in an area with a SIR of 107/100,000 (95 % CI 103–111) for LC in males, that is almost twice the average Italian SIR (i.e. 59/100,000) (Italian National Institute of Tumours 2015; Lombardy Region and Local Health Authority of Brescia 2013; Simonati et al. 2004).

From 1998 to 2013, 3274 cases of LCs were referred to the OHU. Ninety-eight percentage of the patients were hospitalized in Pneumology Unit; approximately 81 % lived in the Brescia Province. About 200 patients/year were referred to the OHU, accounting for more than 85 % of patients assessed at the Pneumology Unit; a few were not evaluated because of health condition, logistic reasons or unwillingness to sustain interview. A total of 1752 cases were filed, since no occupational exposure to lung carcinogens could be identified; 505 required further information; 12 were not interviewed because of poor health conditions or discharge. A total of 1522 patients were fully interviewed. The main characteristics of patients are reported in Table 1. Most of the LC patients were smokers or former smokers. The average number of job tasks in lifetime was 1.6 (range 1–7) for not interviewed cases, 2.8 (range 1–9) for those interviewed.

Among the 1522 LCs fully interviewed, causation was attributed to occupation in 395 cases (26 % of all interviewed patients; 12 % among total cases referred). These cases were all reported to the Local Health Authority; very few LC (just a couple of dozens) were instead reported before the SS.

Figure 1 shows the distribution of occupational LC by activity and carcinogenic exposure.

In addition to LC, other occupational disorders were identified such as 4 asbestosis, 2 pneumoconiosis, 1 systemic sclerosis, 8 urinary bladder cancer, 1 chronic obstructive pulmonary disease, 21 pleural benign asbestos lesions. A total of 28 subjects were already compensated for occupational diseases (23 silicosis, 1 silico-anthracosis, 2 asbestosis, 2 mixed dust pneumoconiosis); in 15 cases, silicosis was discovered during SS.

INAIL data, updated until May 2010 only, show that 240 claims of occupational disease were filed; among the 234 concluded cases, 91 (39 %) were compensated, 143 (61 %) rejected; 6 were under assessment.

Compensated cases included 27 silica workers (20 with silicosis), 15 painters, 10 asbestos workers, 8 truck drivers, 3 electroplaters, 2 steel foundry workers, 2 polycyclic aromatic hydrocarbon-exposed workers, 1 car mechanic, 1 tar sprayer, 1 sulphuric acid-exposed worker, 1 physician exposed to ionizing radiations, 1 rubber worker, 19 with multiple exposures.

A total of 143 cases were rejected by INAIL; motivations for rejections were mainly stated as lack of the exposure to risk, lack of causal association, denial of occupational disease, inadequate clinical or administrative documentation, lack of jurisdiction. A total of 116

Table 1 Caselist distribution by gender, age, smoking habits, histological type and attribution to occupation

Variable of interest	Total caselist		Patients not interviewed		Patients interviewed		Lung cancer not attributed to occupation		Lung cancer attributed to occupation	
	N	%	*N*	%	*N*	%	*N*	%	*N*	%
Cases	3274	100	1752	54	1522	46	1127	74	395	26
Gender										
Males	2823	86	1338	76	1485	98	1098	97	387	98
Females	451	14	414	24	37	2	29	3	8	2
Age (years)										
30–39	26	1	16	1	10	1	5	0.5	5	1
40–49	134	4	85	5	49	3	12	1.1	37	9
50–59	559	17	303	17	256	17	158	14	98	25
60–69	1261	38	640	37	621	40	462	41	159	40
≥ 70	1249	38	670	38	579	38	486	43	93	24
Missing	45	2	38	2	7	1	4	0.4	3	1
Smoking habits										
Current smokers	1250	38	715	41	535	35	405	36	130	33
Former smokers	1478	45	630	36	848	56	613	54	235	59
Non-smokers	546	17	407	23	139	9	109	10	30	8
Histological type										
Adenocarcinoma	1128	35	632	36	496	33	372	33	124	31
Squamous carcinoma	830	25	390	22	440	29	308	27	132	33
Small cell carcinoma	422	13	213	12	209	14	144	13	65	17
Large cell carcinoma	62	2	31	2	31	2	22	2	9	2
'Non-oat'	335	10	193	11	142	9	128	11	14	4
Others[a]	497	15	293	17	204	13	153	14	51	13

[a] Anaplastic/undifferentiated; mixed; not otherwise specified; sarcomatous/pseudosarcomatous; carcinoid/neuroendocrine

cases attributed to occupation were not followed up, neither filed by local INAIL because of their residence outside province or because certificates did not actually reach INAIL. Unfortunately, notwithstanding numerous attempts, it was not possible to get more information from INAIL. Also, it was likely that INAIL did not get some certificate, mainly because: the worker (or ex-worker) has the right not to deliver it; the certificate was not actually given to the worker; workers forget to deliver it to INAIL or loose the certificate, or fear some retaliation.

Among cases rejected by INAIL, 24 subjects (8 % of total LC attributed to occupation) directly approached patronage counselling (helping patients through administrative and medico-legal obligations, as well as legal assistance). Among the 18 cases fully evaluated, occupational aetiology was confirmed in 12 (67 %) and compensated through judicial trial (silica exposure, 5 cases; diesel exhausts, 2 cases; multiple exposures, 4 cases; painter, 1 case); 6 cases were denied (diesel exposure, 3 cases; silica exposure, 2 cases; chromium exposure, 1 case); 6 cases were not concluded. For cases confirmed through judicial process, motivations were mainly based on OHU evaluations. For denied cases, an important role was attributed to smoking habits.

Discussion

The gap among occupational LC epidemiologically expected and those reported and compensated is remarkable and unacceptable from scientific, social and ethical standpoints. Several reasons explain such underestimation.

Physicians play a key role in reporting occupational LC; however, they may fail to recognize work-relatedness, owing to: limited knowledge about occupational cancers and aetiological diagnosis; scientific uncertainties and poor risk assessment data; overweight of confounding factors and the stigma associated to smoking and LC; long latency and changes of exposure patterns over time; lack of clinical specificity of LC; bureaucracy; passive attitude with little

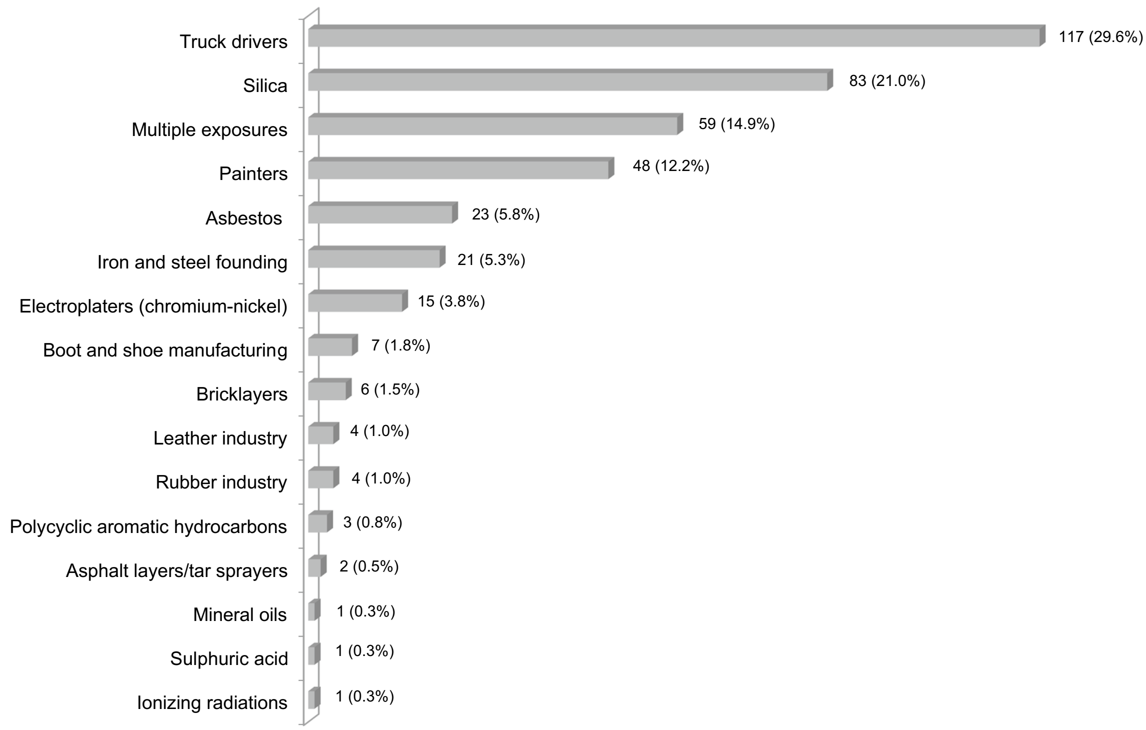

Fig. 1 Occupational lung cancers (*N*, %): distribution by occupational activity, exposure circumstances or carcinogenic agent (single or mixture). Total cases assessed by the OHU: 395

time dedicated to occupational history collection and risk assessment; fear of retaliations. Moreover, workers, who are supposed to file claims, might ignore their rights and opportunities, or avoid to report because of fear of disciplinary actions, denial of opportunities and even job loss, or may not be able to sustain administrative obstacles (compensation may be denied, pays for only a portion of lost wages, long and often unsuccessful procedures). Besides, there might be an attitude to minimize carcinogenic risks by employers, risk assessors, employees and OP as well as a very restrictive approach in adjudication policies of compensation authorities (Ahn and Jeong 2014; Azaroff et al. 2002; Fan et al. 2006; Friedman and Forst 2007; Scarselli et al. 2009; Viau et al. 2008). The combination of these factors leads to underreporting, underestimation and undercompensation of occupational LC. Therefore, actions must be taken to contrast this phenomenon and find lost LC.

In Italy, very few studies have been carried out to search lost cancer, particularly lung and bladder (Albanese et al. 2007; Cosentino et al. 2007), primarily through telephone interviews, evaluation of hospital discharge codes, rarely through direct interview of hospitalized patients; assessed occupational LCs were very few. Similar experiences were carried out in Belgium (Morelle et al. 2014) and in Norway (Slåstad et al. 2014) again, however, with very few cases assessed and limited data. In France, a number of experiences were carried out: a study detected 63 occupational LCs through questionnaires set by an OP, during hospitalization (De Lamberterie et al. 2002); other authors carried out a small research in a Paris hospital; claim for compensation was filed for 32 subjects, mainly for asbestos (Legrand Cattan et al. 2000; Pairon et al. 2006).

Another study (Cellier et al. 2013) developed a questionnaire-based approach to select LC patients for occupational health consultation, and 17 patients were filed for compensation. Other authors (Spreeuwers et al. 2009) suggested criteria for diagnosis and notification, for quality assessment and improvement of registries of occupational diseases. South Korea developed a reporting protocol for occupational LC surveillance; 3353 patients were interviewed: 77, 316 and 314 cases of definite, probable and possible occupational LC were respectively reported, with no collection of smoking habits (Leem et al. 2010).

A peculiar method to find occupational hazards used a case–control approach in Italy (Crosignani et al. 2006). Cases were from hospital records, controls from source populations, and economic sector of the employing company was used as 'exposure'. Many industries at risk were identified, as well as cases of hypothesized occupational origin. However, information on employees in important areas like agriculture, self-employment and the public sector was not available, as well as detailed information about job tasks, therefore preventing from identification of real causal associations.

Italian cancer registries have also contributed to increase the number of referred cases, but only for mesotheliomas and sinonasal cancers; it should also be underlined that such registries do not refer LC patients to OHU or INAIL (Italian National Institute for Prevention and Safety at Work-ISPESL 2008; Italian Workers' Compensation Authority-INAIL 2012). Overall, the above-reported surveys, while unfortunately yielding limited numbers of occupational cancers and collecting limited data for short periods of time and carried out with different purposes, undoubtedly encouraged proactive approaches to reduce undernotification and undercompensation.

As compared to the above-reported studies, our research adopted two main lines of intervention, not followed by other studies:

(a) a systematic approach, searching LC cases where they are diagnosed and treated, taking advantage of the in-house OHU;
(b) attention to aetiological diagnosis, an important moment, considering its consequences according to clinical, epidemiological, preventive, individual and public health standpoints.

Our approach seemed to have several strengths and key points:

(a) high yield; we filed 3274 reports and 1522 evaluations over 16 years, with 395 diagnosis of occupational LC, as compared to the couple of dozen cases reported over 1987–1998, when no search was in-place. Moreover, our caselist is the largest we found in the literature. This testifies that LCs were significantly 'lost' and that they could be 'found' through SS;
(b) better exposures assessment and diagnosis, through high percentage of bedside interview, allowing direct acquisition of occupational history and confounding factors, as well as fresh clinical diagnosis and documentation; this avoided retrospective assessments using hospital discharge codes or routinely collected data, requiring time-consuming acquisition of information, with potential misclassifications;
(c) significant reduction of underreporting, due to proactive approach through OHU supervision and promotion of awareness of clinicians, now prone to notify;
(d) contribution to preventive actions in workplaces. In selected circumstances, especially for those LC patients still working we had: better risk assessment, risk removal; focussed health surveillance, counselling for fitness and return to work; workplace inspections. Such interventions were performed through enhanced cooperation with plant OP, Local Health Authorities, employers, risk assessors, other occupational health professionals, trade unions;
(e) individual case management, counselling and follow-up of compensation and litigation; a greater number of workers were cared for as compared to other types of occupational cancers search. Also, only a handful of LCs were compensated during 1987–1998, whereas compensated LCs were 91 out of 234 evaluated (these data do not include all assessed cases, since information on compensation was available for a limited period of time); our percentage of compensations (39 %) was greater than that recorded at national (average compensation rate of 23 %, all diseases included) and local level (14 % of compensations, all diseases included); the great majority (i.e. 12 out of 14, 86 %) of occupational LCs compensated by INAIL during 2001–2005 in the Lombardy Region were from the province of Brescia (INCA CGIL Patronage 2006; Italian Workers' Compensation Authority-INAIL 1998–2013).

    Given the purpose of the present work, we of course did not intervene on the INAIL compensation scheme, which follows procedures for causal attribution different from ours (a striking example being the difference for truck drivers, see Fig. 1 and text), INAIL pursuing in fact different purposes using appraisal systems (Scarselli et al. 2009) with which we disagree on a strict scientific viewpoint; we are not aware of any change in attribution process along SS period. However, in our opinion, this greater percentage of compensation was likely due to application of sound occupational health methodology, which enabled collection of relevant documentation to sustain causal association, discussion of cases and reference to scientific literature, enabling easier evaluation of cases and burden of proof; for cases rejected from INAIL, judgement appeal carried out by patronages led to compensation on the ground of the same documentation produced from OP during first diagnostic evaluation (67 % cases);

(f) on the public health authorities and judiciary sides, information was given by the OHU to enable solution of specific cases and to help decide whether to proceed with autopsy, inspections or other judicial activities;
(g) teaching opportunities for medical students and residents in Occupational Health;
(h) adoption of such methodology in other OHU of Lombardy Region, with a relevant increase of occupational LC cases diagnosed and referred to Health Authority (in 6 years 2315 cases evaluated, 700 attributed to occupation).

Regarding aetiology, our findings agree with the current literature, for type, duration of exposures and latency (Ahn and Jeong 2014; Baan et al. 2009; Gamble et al. 2012; Guha et al. 2010; International Agency for Research on Cancer-IARC 2013; Olsson et al. 2011; Rota et al. 2014; Steenland et al. 1998; World Health Organization-WHO and International Agency for Research on Cancer-IARC 2014).

The percentage of occupational LC in our caselist was about 26 % (1 out of 4 cases when fully evaluated by OP after his questionnaire selection) and about 12 % (1 out of 10) within the total LC cases referred to the OHU; these percentages are coherent with figures expected according to epidemiological estimates on attributable risk (De Matteis et al. 2008, 2012; Doll and Peto 1981; Driscoll et al. 2005; Straif 2008; World Health Organization-WHO 2009).

Certain limitations of our research should also be acknowledged.

Some underestimation still persists: SS could not reach all LC diagnosed in the area, since some cases were admitted to other hospitals; also, the number of incident LC cases assessed at the Pneumology Unit is estimated around 35–40 % of those expected according to local epidemiological data; moreover, some patients did not request patronage counselling, and some physicians did not report cases to authorities.

Occupational history collection and reconstruction of causal relationship might have been prone to some misclassifications. For example, exposures, technologies, tools, materials could have changed over decades. Moreover, recall from each patient could be difficult in some circumstances, especially considering clinical conditions. To overcome these difficulties, different sources of information were used from companies, Health Authorities, historical OHU files.

As for confounding factors, most LCs were current or former smokers. Nonetheless, reliable collection of smoking habits enables satisfactory control of potential confounding from smoking; moreover, even considering highest grade of confounding, risk attributable to work still persists. In addition, synergistic effect of tobacco smoking in LC genesis is well known in exposure circumstances involving asbestos or polycyclic aromatic hydrocarbons, and methods are available to account for bias due to confounding by smoking in occupational studies (Richardson 2010).

Conclusions

Systematic search of occupational LC cases in a hospital setting in Northern Italy appeared effective and efficient. Within the framework of the principles of Occupational Health, a remarkable number of occupational LC cases were found that otherwise would have been lost. The process of aetiological diagnosis in the context of SS is highly motivated, rich of scientific advantages and practical implications, not to mention its ethical and social aspects. SS is feasible, accountable and could be applicable on wider scale, e.g. for other cancers with a remarkable occupational attributable fraction (e.g. sinonasal cancers or mesotheliomas, bladder cancer, haemo-lymphopoietic system tumours). It should be a strategic activity of Occupational Health Institutions based in hospital and community settings.

In conclusion, professional skills and competence of the qualified, motivated OP, combined with evidence-based approach and SS, and within the framework of a multidisciplinary collaboration with, for example, pneumology, thoracic surgery, pathology, radiology, oncology and occupational medicine, could yield aetiological diagnosis for LC on a large scale, giving a fundamental contribution to reduce underreporting and underestimation of occupational LC.

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Compliance with ethical standard

Conflict of interest The Authors declare that they have no conflict of interest.

Ethical standard All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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