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Estimating the Total Number of Newly-Recognized Silicosis Cases in the United States

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Background *The US employer-based surveillance system for documenting occupational injuries and illnesses undercounts chronic diseases. We suggest a method to estimate the number of individuals who are newly-recognized with silicosis each year in the United States.*

Methods *Data from US death certificates, the Michigan state-based surveillance system, and capture–recapture analysis were used to calculate national estimates of silicosis.*

Results *From 1987 to 1996, 2,787 deaths occurred in the United States where silicosis was mentioned on the death certificates. During the same period, in Michigan 77% of death certificates with a mention of silicosis were confirmed as silicosis-related deaths and the ratio of the number of living to deceased confirmed silicosis cases was 6.44. The proportion of confirmed silicosis deaths, the ratio of the living to deceased silicosis cases and capture–recapture analysis from the Michigan surveillance system, were used to estimate that there were 3,600–7,300 cases per year of silicosis in the United States from 1987 to 1996.*

Conclusions *Our estimate of the annual number of newly-recognized silicosis cases is significantly larger than the estimate from the employer-based reporting system used for counting occupational disease in the United States. This employer-based surveillance system is inadequate for determining the frequency of occupational disease. Our analysis which combines a readily-available and relatively inexpensive national administrative database (i.e., death certificates) with a more costly state-based active surveillance system is a cost-effective model that could be used to provide better estimates of a number of different occupational diseases. Accurate estimates of occupational illnesses are essential to both determine temporal trends and evaluate efforts to prevent silicosis.* Am. J. Ind. Med. 44:141–147, 2003. © *2003 Wiley-Liss, Inc.*

KEY WORDS: disease surveillance; epidemiology; pneumoconiosis; silicosis

INTRODUCTION

The United States relies on an employer-based surveillance system for counting occupational injuries and illnesses, which is administered by the U.S. Department of Labor Bureau of Labor Statistics (BLS). This system is known to markedly undercount chronic diseases [Committee on National Statistics, 1987]. Windau et al. [1991] documented this limitation for the pneumoconioses; four states (California, New Jersey, New York, and Wisconsin) identified 2,910 individuals with pneumoconiosis in 1985 while only 1,700 individuals were reported in the BLS statistics for the entire country in that same year.

The National Institute for Occupational Safety and Health (NIOSH) has compiled its own data on occupational respiratory disease [NIOSH, 1999]. NIOSH has used nationally available databases, death certificates, and hospital discharge data without confirmation of cases in its record

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compilation of pneumoconiosis. Administrative databases by themselves, although available nationwide, have limitations in terms of completeness and accuracy. It is well known that cause of death recording and coding on death certificates are often inaccurate. This inaccuracy can lead to overestimates due to false positives (i.e., the mistaken identification of noncases) and underestimates due to false negatives (i.e., the missing of true cases). The broad scope of the nationwide data makes it difficult or impossible to directly address these problems. NIOSH has also provided approximately 15 states with funding to develop occupational disease surveillance systems for a limited number of conditions. This program is called the Sentinel Event Notification Systems for Occupational Risks (SENSOR). These state-based programs are designed to meet accepted standards for public health surveillance, including case definitions, procedures for case confirmation, and representative data sources. What the state-based programs lack in nationwide coverage they compensate for, with more complete case identification and greater detail about individual cases.

Michigan is one of seven states that has received funding to conduct silicosis surveillance as part of the SENSOR program [Maxfield et al., 1997]. Surveillance for silicosis in Michigan is based on multiple data sources and has been in operation for 14 years [Rosenman et al., 1997].

The goal of the current study was to derive national estimates of the total number of newly-recognized silicosis cases in the United States. This estimate was achieved by combining national mortality data with specific information learned from silicosis surveillance in Michigan. The steps involved included correcting for the overestimation of deceased silicosis cases, estimating the number of newly-recognized living silicosis cases based on the number of deceased cases, and correcting for the underestimation of the number of living silicosis cases identified by surveillance using capture–recapture analysis. This methodology could be applied to other occupational diseases where states have developed sophisticated surveillance systems.

METHODS

Details of the Michigan surveillance system for silicosis have previously been described [Rosenman et al., 1997]. It was initiated in 1987 with financial assistance from NIOSH. Cases of silicosis were identified through: (a) death certificates; (b) reports from physicians; (c) workers' compensation (WC) claims awarded by the Michigan Silicosis, Dust Disease and Logging Industry Compensation Fund; and (d) reports from hospitals. Reporting of all known or suspected occupational diseases by hospitals and physicians was required under Part 56 of Public Act 368 of 1978.

The criteria used to confirm that a person had silicosis was: (a) a history of exposure to silica; and (b) a chest radiograph interpretation showing rounded opacities of 1/0 or greater profusion as per the International Labor Office classification system for pneumoconiosis, or (c) a biopsy report of lung tissue showing fibrosis consistent with silicosis [Maxfield et al., 1997]. Individuals with 10 or more years of underground coal mining experience and less than 3 years of silica exposure were considered to have coal workers' pneumoconiosis and not silicosis. History of exposure was obtained from personal interviews, interviews of next-of-kin, and medical records. Chest X-rays were reviewed by a physician (K.R.) who was a NIOSH-certified ''B-reader'' and therefore had special training and accreditation to interpret chest radiographs for the pneumoconioses, including silicosis. Twenty-six percent had progressive massive fibrosis, 32% had advanced simple silicosis (category 2 or 3), 39% had simple silicosis (category 1), and 3% had biopsy evidence with a normal chest X-ray (category 0).

Estimation of the Total Number of Newly-Recognized Silicosis Cases in the United States

NIOSH obtained the public-use multiple cause of death data file from the National Center for Health Statistics to compile its report on the total number of deaths from silicosis in the United States [NIOSH, 1999]. Both underlying and contributing causes of death were used to identify 2,787 cases with silicosis, during the 10-year period, 1987 through 1996. Starting with these cases, four steps were taken to yield an estimate of the total number of newly-recognized silicosis cases in the US, from 1987 to 1996.

Step 1: Estimate the national number of deaths with a confirmed diagnosis of silicosis

For the years 1987–1996, all Michigan death certificates with silicosis as either the underlying or contributing cause of death were identified through the Michigan surveillance system. By applying the diagnostic criteria outlined above, we determined what proportion of the Michigan silicosis cases identified by death certificate had a confirmed diagnosis of silicosis. Next, the national number of silicosis deaths based on death certificates during 1987–1996 was multiplied by the proportion of the deceased Michigan silicosis cases with a confirmed diagnosis in order to estimate the number of confirmed silicosis deaths in the United States.

In the NIOSH compilation of death certificates which mentioned silicosis for the years 1987–1996, the number of deaths identified in Michigan was 130 [NIOSH, 1999]. The number of cases where silicosis was mentioned on the death certificate as identified by the Michigan surveillance system was 110. The difference is presumed to be secondary to differences between coding by the National Center for Health Statistics and that done by nosologists at the Michigan

Department of Community Health. The 110 number from the Michigan surveillance system was used in the calculations.

Step 2: Estimate the number of newly-recognized living silicosis cases who would have been identified by nationwide surveillance

For the same 10 years, 1987–1996, we identified all living individuals in Michigan with silicosis who were reported by one of the non-death-certificate sources (i.e., physicians, WC, and hospitals) and had not died during the 10-year period with a mention of silicosis on their death certificate. These living cases were confirmed using the same criteria that were applied to deceased cases that had been identified by a death certificate. Based on the confirmed silicosis cases from Michigan, the ratio of the number of living to deceased cases was calculated. This Michigan ratio was then multiplied by the estimated national number of deaths with a confirmed diagnosis of silicosis to obtain an estimate of the national number of newly-recognized living silicosis cases that would have been identified if nationwide surveillance had been conducted.

Step 3: Estimate the total national number of living silicosis cases, including those that would have been missed by surveillance

The Michigan surveillance data were used to perform capture–recapture analysis to estimate the total number of newly-recognized living cases of silicosis, including those who were missed by the state-based surveillance system [Hook and Regal, 1995]. One of the assumptions of the analysis is that candidates for identification have an equal chance of being identified by any one source. Since living cases were not candidates for identification by death certificates, this source was not included in the capture–recapture analysis, and the analysis was limited to confirmed silicosis cases that were not identified by death certificate. The three sources of case included in the analysis were physician reports (PR), WC claims, and hospital reports (HR). Log-linear regression models were fit to control for potential interactions between the three sources while estimating the total number of confirmed silicosis cases. Eight hierarchical models were fit: all sources independent (i.e., no interactions); three models with one pair-wise interaction; three models with two pair-wise interactions; and a saturated model with three pair-wise interactions. Log-linear regression models were fit using the SAS Catmod procedure on a personal computer [SAS, 1989]. Confidence intervals were calculated for each model-specific estimate of cases using a goodness-of-fit approach developed by Hook and Regal [1995].

With the uncertainties inherent in capture–recapture analysis, it is prudent to report a range of estimates. The estimate from the saturated model is suspect since the model is the most complex and the confidence interval is the widest. Two information criteria based on the likelihood ratio statistic (G^2) were computed for each model and used to judge which of the other seven log-linear models provided optimal estimates. The Akaike information criterion (AIC) equals $G^2 - 2$ (d.f.), where d.f. is the degrees of freedom of the model [Hook and Regal, 1995]. The formula for the Bayesian information criterion (BIC) is $G^2 - (\log N_{obs}/2\pi)$ (d.f.), where N_{obs} is the number of observed cases. The preferable models have the lowest information criteria [Hook and Regal, 1995]. The range of estimates of cases was based on results from the three models with the lowest AIC and the three models with the lowest BIC.

The results of the capture–recapture analysis represent the estimated total number of newly-recognized living silicosis cases in Michigan, including those that were missed by surveillance. A ratio was calculated by dividing this estimate by the number of confirmed living cases actually reported to the Michigan surveillance systems. This ratio was then multiplied by the national estimate of newly-recognized living silicosis cases from Step 2 to arrive at the estimated total national number of newly-recognized living silicosis cases.

Step 4: Estimate the total national number of newly-recognized silicosis cases

This was accomplished by adding the number of deceased cases from Step 1 and the number of living cases from Step 3.

RESULTS

Table I summarizes the four steps in the calculations used to arrive at the national estimate of newly-recognized silicosis cases in the United States.

Step 1

NIOSH reported 2,787 deaths in the United States during 1987–1996 (i.e., an average of 279 annually) where silicosis was mentioned on the death certificate. During this same period, Michigan had 110 deaths with silicosis mentioned on the death certificate of which 85 (77.27%) had a confirmed diagnosis. Non-confirmed cases typically had other pneumoconioses such as asbestosis or coal workers' pneumoconiosis. Multiplying $2{,}787 \times 0.7727$, we estimated there were 2,154 confirmed silicosis deaths in the United States during 1987–1996, or an average of 215 deceased silicosis cases per year.

TABLE I. Calculations Used to Estimate Total Number of Newly-Recognized Silicosis Cases, United States, 1987–1996

Reason for calculation	Description of calculation	Numbers
Step 1: Estimate national no. of confirmed silicosis-related deaths	(No. of death certificates which mentioned silicosis, USA) × (proportion of confirmed silicosis-related deaths, Michigan) = Estimated no. of confirmed silicosis-related deaths, USA	2,787 × 0.7727 = 2,154
Step 2: Estimate national no. of living silicosis cases who would have been identified by surveillance	(Estimated no. of confirmed silicosis-related deaths, USA) × (ratio of living to deceased silicosis cases, Michigan) = Estimated no. of surveillance-identified living silicosis cases, USA	2,154 × 6.44 = 13,872
Step 3: Estimate national no. of living silicosis cases, including those who would have been missed by surveillance	(Estimated no. of surveillance-identified living silicosis cases, USA) × (ratio of total no. of cases to surveillance cases, Michigan) = Estimated total no. of newly-recognized living silicosis cases, USA	13,872 × 2.45 = 33,986 to 13,872 × 5.12 = 71,025
Step 4: Estimate national no. of newly-recognized living and deceased silicosis cases	(Estimated no. of confirmed silicosis-related deaths, USA) + (estimated total no. of newly-recognized living silicosis cases, USA) = Estimated total no. of newly-recognized silicosis cases, USA	2,154 + 33,986 = 36,140 to 2,154 + 71,025 = 73,179

Step 2

A total of 632 people with confirmed silicosis were reported to the Michigan surveillance program during 1987–1996. Of that number, 85 were identified by death certificate, leaving 547 who were reported by at least one of the non-death-certificate sources (i.e., physicians, WC claims, hospitals). The ratio of the number of living confirmed silicosis cases to the number of deceased confirmed silicosis cases in Michigan from 1987 to 1996 was 547 divided by 85 or 6.44. Multiplying this ratio by the estimated number of national deaths (2,154) yielded a national estimate of 13,872 newly-recognized living silicosis cases or an average of 1,387 per year. This is the estimated number of living silicosis cases that would have been reported if there had been a nationwide surveillance system for silicosis.

Step 3

The reporting sources for the 547 non-death-certificate cases of silicosis are presented in Table II. Hospitals provided the largest number of reports with 407, followed by 111 from physicians, and 84 from WC. The percentage of cases also reported by at least one of the other sources was 13% (51/407) for HR, 23% (26/111) for PR, and 37% (31/84) for WC. Most of the overlap occurred between the HR and each of the other sources: 24 PR and HR; 29 WC and HR. At the same time, there were only four cases identified by both PR and WC, and only two by all three sources.

The estimates of the total number of non-death-certificate cases from the log-linear models are presented in Table III. With the information criteria AIC and BIC, lower values indicate more favorable models. The model with the

TABLE II. Distribution of 547 Living Cases of Silicosis in Michigan, 1987–1996, by Three Reporting Sources*

	Physician reports (PR)			
	Yes		No	
	Workers' compensation		Workers' compensation	
	Yes	No	Yes	No
Hospital reports (HR)				
Yes	2	22	27	356
No	2	85	53	0

*Does not include the 85 cases with confirmed silicosis that were identified by death certificate.

TABLE III. Three-Source Estimates of the Total Number (N) of Living Silicosis Cases in Michigan During 1987–1996, Based on Capture–Recapture Analysis

Model[a]	d.f.[b]	G^{2b}	*P*	AIC[b]	BIC[b]	N	95% CI
Independence	3	6.0	0.11	−0.04	−7.4	1,569	1,282–1,978
PR–WC	2	5.1	0.08	1.1	−3.8	1,524	1,243–1,932
PR–HR	2	3.6	0.17	−0.4	−5.4	1,339	1,049–1,808
WC–HR	2	0.6	0.75	−3.4	−8.4	1,972	1,465–2,825
PR–WC, PR–HR	1	1.6	0.20	−0.4	−2.8	1,246	981–1,689
PR–WC, WC–HR	1	0.42	0.51	−1.6	−4.0	1,922	1,409–2,825
PR–HR, WC–HR	1	0.05	0.82	−2.0	−4.4	2,800	1,231–14,440
Saturated	0	0	0	0	0	3,247	843–25,217

[a]The models are hierarchical, with lower-order terms for the three sources included in each model. Abbreviations for sources of cases: PR, physician report; WC, workers' compensation; HR, hospital report.
[b]d.f., degrees of freedom; G^2, likelihood ratio statistic; AIC, Akaike information criterion; BIC, Bayesian information criterion.

one interaction WC–HR and an estimate of 1,972 (95% CI 1,465–2,825) living silicosis cases had the lowest value for both AIC and BIC. A pair of two-interaction-term models (i.e., PR–WC, WC–HR and PR–HR, WC–HR) had the next two lowest AIC values, while the independence model (i.e., no interactions) and the model with the one interaction PR–HR had the next two lowest BIC values (Table III). Based on these five models defined by the three lowest AIC and BIC values, the estimates of the total number of living silicosis cases ranged from 1,339 to 2,800. The ratios of these minimum and maximum values to the 547 observed living cases in Michigan were 2.45 and 5.12, respectively. Multiplying these ratios by 13,872 cases yielded nationwide estimates ranging from 33,986 to 71,025 of new-recognized living silicosis cases for the years 1987–1996.

Step 4

When the figures from Step 3 are added to the estimated number of deceased cases from Step 1, the estimated total number of newly-recognized cases of silicosis for the United States ranged from 36,140 to 73,179, or an average number of from 3,600 to 7,300 per year for 1987–1996.

DISCUSSION

Accurate surveillance systems for work-related injuries and illnesses are important for both guiding decision making about the percentage of public health resources that should be allocated to occupational health and safety in comparison to other public health issues, as well as prioritizing and evaluating occupational public health and enforcement activity. Current national statistics for silicosis either based on death certificate data or the BLS employer-based sampling markedly undercount the actual number of individuals being diagnosed with silicosis.

From 1987 to 1996 in the United States there was an average of 279 death certificates per year with silicosis either as the underlying or contributing cause of death. As has been demonstrated with injury surveillance, there is a hierarchy of outcome measures with fatalities being at the top of the pyramid and only representing a very small percentage of possible adverse outcomes. For the same years, 1987–1996, the BLS employer-based sampling system estimated 2,700–3,500 individuals per year with all types of pneumoconiosis. We estimate that for silicosis alone there were on average from 3,600 to 7,300 individuals identified with this disease each year during 1987–1996 (Table IV).

The current employer-based surveillance system is an annual survey based on a sampling scheme that covers only part of the workforce. Self-employed workers, government employees from 27 states, and farms with less than 11 employees are not included. In general, employers are unaware of chronic diseases such as silicosis, which may not develop and/or be diagnosed until years after employment has ceased. Since the majority of workers with silicosis never apply for WC, even if the individual is continuing to work, the employer may be unaware that an employee has been diagnosed with silicosis [Stanbury et al., 1995; Rosenman et al., 1997]. Additionally, there is a potential disincentive to

TABLE IV. Comparison of Annual Estimates of Silicosis in the United States Based on Differing Data Sources

Death certificates [NIOSH, 1999]	279
Bureau of Labor Statistics	2,700–3,500[a]
Capture/recapture analysis based on Ohio data [Windau et al., 1991]	4,900
Our analyses	3,600–7,300

[a] This is an estimate for all types of pneumoconiosis including asbestosis and coal worker's pneumoconiosis as well as silicosis.

employers to record and report these chronic diseases, since such reporting may increase enforcement inspections or affect compensation claim filing.

There are several potential limitations of the estimates presented in this study. From Step 1 of the calculations (see Table I), it might not be accurate to assume that the percentage of Michigan death certificates excluded (i.e., 23%) because they did not meet the criteria for a confirmed silicosis case is the same across the nation. Individuals without rounded opacities on their chest radiograph, as well as individuals with more than 10 years of underground coal mining experience and less than 3 years of silica exposure, did not meet the criteria for a confirmed silicosis case. In states with a more extensive history of shipyards and asbestos exposure or in coal mining states, the percentage of non-confirmed death certificates might be greater than in Michigan. This would occur because the number of individuals with asbestosis or coal worker pneumoconiosis will be increased in those states and given a similar proportion of misdiagnoses the number of unconfirmed silicosis cases that really have another type of pneumoconiosis would increase. Alternatively, one could argue that we excluded people with true silicosis because our criteria for confirming a case of silicosis excluded individuals whose only radiographic changes were linear changes, and therefore the percentage of non-confirmed death certificates for silicosis should be lower. Although we have data to support our criteria regarding the use of rounded opacities [Rosenman et al., 1996; Rosenman and Reilly, 1998], other studies have not made this distinction between types of pneumoconiosis based on opacity shape.

From Step 2 of the calculations, the ratio of newly recognized confirmed cases of silicosis that are living versus those who are dead might not be the same nationwide as in Michigan. For example, physician awareness of silicosis in the state of Michigan may be greater or less than in other states. Less awareness among health care providers in other states may cause physicians to be less likely to accurately complete the death certificate, which would decrease the frequency with which silicosis appears on the death certificate and increase the living to deceased ratio used in the calculation and increase the national estimate. Silicosis was only mentioned on 85 of 254 (33.5%) of the death certificates of confirmed cases of silicosis who were reported to the Michigan surveillance system while they were living and subsequently died. On the other hand, decreased health care provider awareness of silicosis would make it less likely that less severe cases (living cases) of silicosis were recognized. This would decrease the living to deceased ratio used in the calculations and decrease the overall estimate.

The fact that Michigan is more industrialized than some states does not effect the final national estimate. The total number of individuals with silicosis in Michigan does not effect our calculations. Our calculations are dependent on the ratio between living and dead silicotics which should not be effected by the level of industrialization in Michigan. The only way that the degree of industrialization in Michigan would effect our estimate would be if as described above, industrialization caused Michigan physicians to have a greater awareness of silicosis. We have no evidence to suggest that Michigan physicians have more or less awareness of silicosis than physicians in other states. However, silicosis is still a rare disease in Michigan and we doubt that Michigan physicians are more knowledgeable about this diagnosis than physicians in other states.

In Step 3 of the estimation process, we assumed that the capture–recapture analysis as applied to the Michigan data is valid, and that the extent of under-estimation of living silicosis cases in Michigan is the same nationwide. Capture–recapture analysis was used to estimate the number of cases that were missed by surveillance. Guidelines for the implementation of capture–recapture analysis continue to be discussed in the professional literature [Hook and Regal, 2000; Tilling, 2001], and it is important to bear in mind that the number of cases missed by surveillance as determined by this analysis is indeed an estimate subject to error. For example, the validity of capture–recapture analysis is based on a number of assumptions, including: (a) the study population has no losses or entries during the period of investigation; (b) all cases in the population have the same probability of identification in any one source; and (c) ascertainment in any two sources are independent events. These assumptions are not fully met in most epidemiologic studies. Concerning the first assumption, there was movement in and out of Michigan, with a net increase in population from approximately 9,205,000 in 1987 to 9,734,000 in 1996. This increase of about 6% over 10 years is a relatively small change in the size of the study population. The second assumption that all cases have the same probability of identification in any one source stimulated us to exclude one source of cases. In particular, death certificates were not used as a source of cases since living cases were not candidates for identification by this source. Violation of the second assumption was still possible, since severe cases were probably preferentially reported by one of the three sources (hospital discharge data) that were used in the analysis. With respect to the third assumption, interaction terms in the log-linear models allowed us to account for possible violations of independence between the different reporting sources.

Capture–recapture analysis was also used with SENSOR surveillance data in Ohio to estimate the total number of silicosis cases in Ohio, both living and deceased, identified during 1989–1995 [Geidenberger and Socie, 1997]. The best estimates from that analysis revealed that the total number of silicosis cases was about 3.03 to 3.18 times the number of cases identified by surveillance. Using the smaller of the two Ohio estimates to multiply our national surveillance estimate of 16,026 (i.e., 2,154 deceased from Step 1 plus 13,872 living from Step 2) yielded a national estimate of 48,559, or an

average of approximately 4,900 newly-recognized deceased and living cases of silicosis per year. This estimate is within the range of estimates (i.e., 3,600 to 7,300) from the current analysis.

By combining a readily available and relatively inexpensive national administrative database (death certificates) with a more costly state-based active surveillance system we have been able to calculate national disease estimates. Our estimates are more comprehensive than the current employer-based occupational injury and illness surveillance system, which markedly undercounts the silicosis disease burden. The methodology we have used is a model that could be used for other occupational injuries and illnesses. If costly multiple source surveillance is limited to a few states, the comparison of these surveillance data from multiple sources in these sample states could be used to check the completeness and quality of cases identified in the same states by inexpensive administrative database(s). The differences in the costly multiple source and inexpensive administrative surveillance in these sample states could then be combined with national administrative databases to derive more accurate rates than the current employer-based system. Further analyses with other occupational injuries and illnesses similar to what we have done with silicosis are currently underway in Michigan.

Additional work is needed to determine both the optimum number and sample of states with representative industries where active surveillance should be performed in order to generate the most accurate and cost-effective national estimates. The major assumptions used in our methodology are the percentage of silicosis deaths that are confirmed cases and the ratio of living to dead silicotics. The selection of which states to use to generate national estimates is dependent on including states with stable surveillance systems that have sufficient number of individuals with silicosis. This criteria will minimize errors which may be introduced from variation in the functioning of the surveillance system and that are inherent with the use of small numbers. The inclusion of more industrialized states in the sample states will not cause an overestimate in the final estimate since it would not effect the ratio of living to dead with silicosis unless physicians in these states are more likely to recognize cases of silicosis at an earlier stage while the individual is alive. Further work on the ratio of living to dead silicosis cases in different states would be needed to clarify this possibility. If the ratio were not a factor, including more industrialized states with more disease would increase the stability of the national estimates. It is likely that both the number of states and sample of states will vary by disease condition given the differences in incidence of particular conditions and geographic variation of particular industries.

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