




Cancer-related mortality among people with intellectual disabilities: A nationwide population-based cohort study

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BACKGROUND: Concerns have been raised about access to cancer screening and the timely receipt of cancer care for people with an intellectual disability (ID). However, knowledge about cancer mortality as a potential consequence of these disparities is still limited. This study, therefore, compared cancer-related mortality patterns between people with and without ID. **METHODS:** A historical cohort study (2015-2019) linked the Dutch adult population (approximately 12 million people with an ID prevalence of 1.45%) and mortality registries. Cancer-related mortality was identified by the underlying cause of death (according to the chapter on neoplasms in the *International Classification of Diseases, Tenth Revision*). Observed mortality and calculated age- and sex-standardized mortality ratios (SMRs) with 95% confidence intervals (CIs) were reported. **RESULTS:** There were 11,102 deaths in the ID population (21.7% cancer-related; n = 2408) and 730,405 deaths in the general population (31.2%; n = 228,120) available for analysis. Cancer was noted as the cause of death more often among people with ID in comparison with the general population (SMR, 1.48; 95% CI, 1.42-1.54), particularly in the young age groups. High-mortality cancers included cancers within the national screening program (SMRs, 1.43-1.94), digestive cancers (SMRs, 1.24-2.56), bladder cancer (SMR, 2.07; 95% CI, 1.61-2.54), and cancers of unknown primary (SMR, 2.48; 95% CI, 2.06-2.89). **CONCLUSIONS:** Cancer was reported as the cause of death approximately 1.5 times more often in people with ID compared with the general population. This mortality disparity may indicate adverse effects from inequalities in screening and cancer care experienced by people with ID. **Cancer 2022;128:1267-1274.** © 2021 The Authors. *Cancer* published by Wiley Periodicals LLC on behalf of American Cancer Society This is an open access article under the terms of the Creative Commons Attribution-NonCommercial License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes.

LAY SUMMARY:

- People with an intellectual disability (ID) may find it challenging to participate in cancer screening or to receive timely cancer care.
- To understand potential consequences in terms of mortality, this study compared cancer-related mortality between people with and without ID in the Netherlands.
- Cancer was reported as the cause of death approximately 1.5 times more often among people with ID than others.
- Because large differences were found that were related to screening cancers and cancers for which the primary tumor was unknown, this study's results raise concerns about equality in screening practices and cancer care for people with ID.

KEYWORDS: cancer, cause of death, disparities, intellectual disability, population-based, standardized mortality ratio.

INTRODUCTION

Patterns of cancer mortality are generally well documented and provide insight into the impact of cancer at the population level, the effectiveness of preventive (public health) measures, and the quality of cancer care.^{1,2} Knowledge of cancer mortality among people with an intellectual disability (ID), however, is neither complete nor up to date.^{3,4} Individuals with ID have significant impairments in intelligence and social functioning, which are often caused by genetic mutations, and they account for at least 1.5% of the population in Western countries.⁵⁻⁷ Tumor growth propensities of some ID syndromes could put people with ID at a higher risk of developing certain types of cancer.⁸⁻¹¹ Other risk factors (eg, lifestyle and external factors) are as equally likely to appear among people with ID as among other people, although differences in the age structure of the intellectual disability population (IDpop) with respect to the general population complicate the interpretation of cancer incidence rates.^{12,13}

For their cancer care, people with ID rely in most countries, including the Netherlands, on the same health care systems as people from the general population.¹⁴ This requires care providers to adapt to the specific care needs of people with ID. Moreover, the recognition of cancer symptoms in individuals with ID may be overshadowed by the manifestation of ID traits or asymptomatic presentation of cancer symptoms.^{4,15,16} Combined with a lower likelihood

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of participation in cancer screening, this raises concerns about the accessibility and effectiveness of cancer diagnostics and care for people with ID.¹⁷⁻²¹ Even within health systems that strive for broad accessibility and equitable care, such disparities in cancer care can occur.^{22,23}

For evaluating the overall effectiveness of public health care for people with ID, mortality statistics have become an important source of information.^{24,25} It has, among others, identified elevated risks for premature and potentially avoidable mortality in comparison with the general population.²⁴⁻²⁶ To improve our insight into the impact of cancer among people with ID, this study aimed to characterize cancer-related mortality in Dutch individuals with ID in comparison with the general population.

MATERIALS AND METHODS

Setting and Design

This historical cohort study (2015-2019) used administrative data linked at the individual level with nonpublic microdata from Statistics Netherlands. Upon request, these microdata are accessible for statistical and scientific research. We combined databases containing personal information (date of birth and sex), information on ID-related supportive care use, and mortality statistics (date and underlying cause of death). Under Dutch law, large-scale anonymous database studies and posthumous investigations of mortality data are exempted from formal ethical approval. This was confirmed by an assessment of the study protocol by the Radboud University Medical Center institutional ethics committee (2017-3921).

Study Population

Data from the Dutch population register were retrieved for all individuals who were alive and 18 years old or older on January 1, 2015. Individuals with ID were identified through linkage with national databases for chronic care and social benefits. All recipients of ID-related services through any of these systems were classified by the characteristics of their supportive needs because the actual ID diagnoses are not saved in these databases.²⁷ These data on ID-related care needs are available from 2015 onward. Three ID subgroups were generated: 1) those receiving residential ID care (least independent), 2) those receiving nonresidential ID-related chronic care (moderately independent), and 3) a group with mild ID consisting of those receiving social benefits rather than chronic care (fairly independent). This method for identifying individuals with ID in population data has been described in more detail elsewhere²⁷ and has been applied to other studies related

to health and ID before.^{21,28,29} All individuals without ID characterization were analyzed as members of the general population.

Outcome Measure

The main outcome in this study was mortality, which consisted of the date of death and the underlying cause coded according to the *International Classification of Diseases, Tenth Revision (ICD-10)*. Cancer-specific mortality was defined according to the chapter on neoplasms (codes C00-D48; see Supporting Table 1). Within the chapter on neoplasms, we categorized according to the 18 predefined groups (eg, “malignant neoplasms of lip, oral cavity, and pharynx” [C00-C14]) and reported individual cancer types by their 3-position *ICD-10* code (eg, “C15–malignant neoplasm of esophagus”).

Statistical Analysis

We calculated the age at death by subtracting the date of birth from the date of death. Demographics were presented as frequencies (with percentages) or means (with standard deviations). We reported observed deaths for both groups, and we calculated expected deaths for each sex and 10-year age group in the IDpop on the basis of mortality in the same group from the general population. Age- and sex-standardized mortality ratios (SMRs) were then generated by the division of observed mortality by expected mortality, and they were presented with 95% confidence intervals (CIs). SMRs higher than 1.0 indicated increased mortality risks for the ID population, and SMRs less than 1.0 indicated lower risks. Analyses were conducted for overall mortality, cancer-related mortality, and cancer type-specific mortality for the most common causes. Data quality checks and missing data analysis were conducted at Statistics Netherlands before the data were made available for further analysis. Analyses were conducted with SPSS 25.

RESULTS

Demographics and Overall Mortality

Through linking databases, 187,149 adults with an ID (the IDpop) were identified (ID prevalence, 1.45%) at the start of follow-up in 2015, and the remaining 12,677,768 individuals without ID were assumed to constitute the general Dutch adult population (the GenPop). The mean age was 8.7 years lower in the IDpop than the GenPop (39.6 vs 48.3 years). The IDpop had more males (57.4%) than the GenPop (48.9%). The largest group in the IDpop consisted of recipients of

residential ID care ($n = 91,064$; 48.7%), and they were followed by people with mild ID ($n = 69,078$; 36.9%) and nonresidential chronic care users ($n = 27,007$; 14.4%). A complete overview of all demographics is presented in Table 1.

During follow-up, 11,102 deaths (all causes) were counted in the IDpop, and 730,405 deaths were counted in the GenPop. The mean age at death was 15.6 years lower in the IDpop than the GenPop (63.2 vs 78.8 years). Mortality was higher in the IDpop than the GenPop (SMR, 2.71; 95% CI, 2.66-2.76). SMRs declined with increasing independence across ID subgroups (Table 1).

Cancer Mortality

Cancer was the most common cause of death in the IDpop (22.4% of all deaths), and it was followed by circulatory (20.2%) and respiratory causes (12.7%); this made the top 3 causes of death in terms of *ICD-10* chapters similar to the GenPop. At the IDpop level, there were 785 more deaths due to cancer during follow-up than expected on the basis of cancer mortality in the GenPop (SMR, 1.48; 95% CI, 1.42-1.54; Table 2). Individuals with ID who died of cancer were 10.7 years younger than individuals who died of cancer in the GenPop (62.9 vs 73.6 years; Table 2).

All cancer types within the national screening program (colon, breast, and cervix) caused more deaths among people with ID than expected on the basis of mortality in the GenPop, with colon cancer causing the most deaths in absolute numbers ($n = 200$; SMR, 1.66; 95% CI, 1.43-1.89) and with cervical cancer causing the largest difference in comparison with GenPop (SMR, 1.94; 95% CI, 1.02-2.86), even though only 17 deaths from this cause appeared during follow-up in the IDpop. Breast cancer fell between them in terms of both absolute and relative mortality ($n = 151$; SMR, 1.43; 95% CI, 1.21-1.66; Table 3). By volume, lung cancer was the most common cancer-related cause of death in the IDpop ($n = 493$; SMR, 1.24; 95% CI, 1.13-1.35). The largest differences in comparison with the GenPop were related to cancers of unknown primary (SMR, 2.48; 95% CI, 2.06-2.89) and other (not colon) digestive organs (SMR, 2.65; 95% CI, 2.06-3.25). Among the most common cancer-related causes of death, prostate cancer did not cause elevated mortality in the IDpop in comparison with the GenPop (SMR, 0.91; 95% CI, 0.70-1.12; Table 3).

DISCUSSION

This study is the first to characterize cancer-related mortality among adults with ID by using population

data. Even though life expectancy is lower for people with ID than the general population, we have shown that cancer is the most prominent cause of death for people with ID as well. Although excess mortality due to cancer was less pronounced in comparison with other causes of death, still almost 800 more people with ID died of cancer during the study period than expected on the basis of cancer-related mortality among people of the same age and sex in the general population. This could be the result of 1) a higher overall incidence of cancer among people with ID; 2) a higher incidence of lethal cancers in particular; or 3) less effective cancer care for people with ID, including prevention and screening policies.

First, we consider the possibility of a higher overall incidence of cancer among people with ID. So far, most studies have reported a lower or similar overall cancer incidence among people with ID in comparison with people without ID.^{3,12,13} Even at older ages, the frequency of cancer diagnoses among people with ID appears to be lower in comparison with the general population.³⁰ Specific ID syndromes may be associated with increased risks for certain cancers. For example, Down syndrome is associated with an increased risk for leukemia, yet it is also associated with a reduced risk for solid tumors.^{8,9,31} Although complete information on all genetic ID syndromes with tumor growth propensities is missing to date, these subpopulations appear too small to explain the differences in mortality detected in this study.

Second, lethal cancers in particular could be more prevalent among people with ID than people without ID. There were relatively many deaths due to cancers of unknown primary in the IDpop, and these cancers are often characterized by the presence of metastases at diagnosis and an absence of targeted therapies.³² There were also more deaths from esophageal cancer and digestive cancers in the IDpop than expected. In particular, for advanced stages of these cancers, survival rates beyond the first year after diagnosis are below 50%, and 5-year survival is rare.³³ Risk factors for these cancers include smoking, substance abuse, and poor nutrition, which are all relatively common in certain ID subgroups.^{34,35} Another risk factor for gastrointestinal malignancies is *Helicobacter pylori* infection, which has been reported to occur twice as often among people with ID in comparison with the general population.^{36,37} Moreover, people with ID could be more susceptible to complications after treatment for gastrointestinal malignancies because of their generally greater likelihood

TABLE 1. Demographics and Overall Mortality (All Causes), 2015-2019

	GenPop (n = 12,677,678)		IDpop (n = 187,149)		SMR		
	Population, No. (%)	Observed Deaths, No. (Rate) ^a	Population, No. (%)	Observed Deaths, No. (Rate) ^a	SMR	95% CI	
Total No.	12,677,768	730,405 (115.2)	187,149	11,102 (118.6)	4093	2.71	2.66-2.76
Sex							
Males	6,196,789 (48.9)	352,224 (113.7)	107,370 (57.4)	6323 (117.8)	2515	2.51	2.45-2.58
Females	6,480,979 (51.1)	378,181 (116.7)	78,779 (42.6)	4779 (119.8)	1578	3.03	2.94-3.11
Age, mean (SD), y	48.3 (17.8)		39.6 (15.6)				
Age at death, mean (SD), y	78.8 (12.7)		63.2 (14.0)				
Age at enrollment							
18-24 y	1,362,047 (10.7)	1962 (2.9)	44,343 (23.7)	282 (12.7)	67	4.21	3.71-4.70
25-34 y	1,931,948 (15.2)	4456 (4.6)	36,615 (19.6)	390 (21.3)	87	4.49	4.05-4.94
35-44 y	2,225,395 (17.6)	12,797 (11.5)	32,514 (17.4)	830 (51.1)	191	4.35	4.05-4.65
45-54 y	2,457,868 (19.4)	39,033 (31.8)	37,109 (19.8)	2371 (127.8)	600	3.95	3.79-4.11
55-64 y	2,104,631 (16.6)	89,895 (85.4)	25,140 (13.4)	3490 (277.6)	1110	3.14	3.04-3.25
65-74 y	1,551,963 (12.2)	166,182 (214.2)	8770 (4.7)	2280 (520.0)	973	2.34	2.25-2.44
≥75 y	1,043,916 (8.2)	416,080 (797.2)	2658 (1.4)	1459 (1097.4)	1065	1.37	1.30-1.44
ID group							
Residential ID	—	—	91,064 (48.7)	7262 (159.5)	2414	3.01	2.94-3.08
Nonresidential	—	—	27,007 (14.4)	1621 (120.0)	632	2.56	2.44-2.69
Mild ID	—	—	69,078 (36.9)	2219 (64.2)	1046	2.12	2.03-2.21

Abbreviations: CI, confidence interval; GenPop, general Dutch adult population; ID, intellectual disability; IDpop, intellectual disability population; SMR, standardized mortality ratio.
^aCrude mortality rate per 10,000 per year.

TABLE 2. Cancer-Related Mortality, 2015-2019

	GenPop (n = 12,677,678)	IDpop (n = 187,149)		SMR	
	Cancer-Related Deaths, No. (Rate) ^a	Cancer-Related Deaths, No. (Rate) ^a	Expected Deaths ^b	SMR	95% CI
Total cancer-related deaths ^c	228,120 (36.0)	2408 (25.7)	1623	1.48	1.42-1.54
Sex					
Males	123,041 (39.7)	1383 (25.8)	1002	1.38	1.31-1.45
Females	105,079 (32.4)	1025 (25.7)	621	1.65	1.55-1.75
Age at death, mean (SD), y	73.6 (11.9)	62.9 (11.6)			
Age at enrollment					
18-24 y	294 (0.4)	23 (1.0)	10	2.38	1.40-3.35
25-34 y	1293 (1.3)	47 (2.6)	24	1.97	1.40-2.53
35-44 y	5419 (4.9)	201 (12.4)	78	2.57	2.22-2.93
45-54 y	20,040 (16.3)	576 (31.0)	300	1.92	1.76-2.08
55-64 y	47,159 (44.8)	866 (68.9)	574	1.51	1.41-1.61
65-74 y	71,843 (92.6)	489 (111.5)	421	1.16	1.06-1.26
≥75 y	82,072 (157.2)	206 (155.0)	216	0.95	0.82-1.08
Neoplasm chapter					
Lip, oral cavity, and pharynx (C00-C14)	3304 (0.5)	50 (0.5)	29	1.72	1.24-2.19
Digestive organs (C15-C26)	67,375 (10.6)	778 (8.3)	488	1.59	1.48-1.71
Respiratory and intrathoracic organs (C30-C39)	52,613 (8.3)	518 (5.5)	410	1.26	1.15-1.37
Bone and articular cartilage (C40-C41)	468 (0.1)	5 (0.1)	5	1.08	0.13-2.02
Melanoma and other malignant neoplasms of skin (C43-C44)	4459 (0.7)	31 (0.3)	36	0.85	0.55-1.15
Mesothelial and soft tissue (C45-C49)	3802 (0.6)	27 (0.3)	29	0.92	0.57-1.27
Breast (C50)	15,508 (2.4)	151 (1.6)	105	1.43	1.21-1.66
Female genital organs (C51-C58)	9519 (2.9)	99 (2.5)	58	1.70	1.37-2.04
Male genital organs (C60-C63)	14,239 (4.6)	85 (1.6)	84	1.01	0.79-1.22
Urinary tract (C64-C68)	13,771 (2.2)	135 (1.4)	94	1.44	1.20-1.69
Eye, brain, and other parts of central nervous system (C69-C72)	5086 (0.8)	62 (0.7)	54	1.15	0.86-1.43
Thyroid and other endocrine glands (C73-C75)	1008 (0.2)	10 (0.1)	8	1.29	0.49-2.08
Ill-defined, secondary, and unspecified sites (C76-C80)	11,041 (1.7)	162 (1.7)	63	2.58	2.18-2.98
Primary of lymphoid, haematopoietic, and related tissue (C81-C96)	17,478 (2.8)	170 (1.8)	115	1.48	1.26-1.70
In situ neoplasms (D00-D09)	—	—	—	—	—
Benign neoplasms (D10-D36)	8445 (1.3)	125 (1.3)	44	2.81	2.32-3.31
Neoplasms of uncertain or unknown behavior (D37-D48)	11,798 (1.9)	104 (1.1)	40	2.57	2.08-3.07

Abbreviations: CI, confidence interval; GenPop, general Dutch adult population; IDpop, intellectual disability population; SMR, standardized mortality ratio.

^aCrude mortality rate per 10,000 per year.

^bExpected counts have been rounded.

^cThe chapter on neoplasms accounted for 32.4% of all deaths in the GenPop and for 22.4% of all deaths in the IDpop, with these being the most common cause of death in both groups. Circulatory and respiratory causes were second and third most common in both groups (GenPop, 26.5% and 8.8%, respectively; IDpop, 20.2% and 12.7%, respectively).

of dysphagia and choking problems.³⁸ So far, no ID-specific treatment recommendations are available for clinicians to weigh these specific risks when they are considering treatment.

A potential third explanation for higher cancer mortality is less effective cancer care for people with ID than for other people. This is substantiated by our earlier finding that Dutch people with ID were given less cancer-related care than matched individuals without ID of the same age and sex.²¹ Other literature suggests that people with ID also participate poorly in screening programs, and this lowers their chances for early detection of cancer types targeted by screening.^{20,39-41} For all cancer types in

the Dutch screening program (colon, cervical, and female breast), this study showed elevated mortality for people with ID in comparison with the general population. This underlines international concerns about access to and participation in population screening and shows the need to investigate this further.

The age disparity in cancer mortality between people with ID and the GenPop could be an indicator for late diagnoses, less extensive treatments, and suboptimal overall disease management. However, death at a younger age for people with ID should also be interpreted with respect to an earlier onset of aging and the occurrence of age-related diseases at a younger age in

TABLE 3. Most Common Cancer Type-Specific Causes, 2015-2019

	GenPop (n = 12,677,678)	IDpop (n = 187,149)		SMR	
	Cancer-Related Deaths, No. (Rate) ^a	Cancer-Related Deaths, No. (Rate) ^a	Expected Deaths ^b	SMR	95% CI
Cancer types with screening ^c					
Colon (C18)	18,298 (2.9)	200 (2.1)	121	1.66	1.43-1.89
Breast (C50)	15,508 (2.4)	151 (1.6)	105	1.43	1.21-1.66
Cervix uteri (C53)	2064 (0.3)	17 (0.2)	9	1.94	1.02-2.86
Cancer types without screening					
Bronchus and lung (C34)	51,145 (8.1)	493 (5.3)	398	1.24	1.13-1.35
Cancer without specification of site (C80)	9585 (1.5)	138 (1.5)	56	2.48	2.06-2.89
Pancreas (C25)	14,047 (2.2)	128 (1.4)	103	1.24	1.03-1.46
Esophagus (C15)	9345 (1.5)	126 (1.3)	81	1.56	1.29-1.84
Other (ill-defined) digestive organs (C26)	5091 (0.8)	77 (0.8)	29	2.65	2.06-3.25
Bladder (C67)	6160 (1.0)	76 (0.8)	37	2.07	1.61-2.54
Rectum (C20)	5664 (0.9)	74 (0.8)	43	1.71	1.32-2.10
Prostate (C61)	13,955 (2.2)	71 (0.8)	81	0.91	0.70-1.12

Abbreviations: CI, confidence interval; GenPop, general Dutch adult population; IDpop, intellectual disability population; SMR, standardized mortality ratio.

^aCrude mortality rate per 10,000 per year.

^bExpected counts have been rounded.

^cAccording to the active population screening program in the Netherlands during the study period (2015-2019). Colon cancer screening was begun in 2014 and fully implemented in 2019. Prostate cancer, for example, also has screening opportunities (prostate-specific antigen testing) but is not part of population screening in the Netherlands.

comparison with people without ID.^{42,43} Because age is a strong predictor for developing cancer, early aging could then potentially also cause younger deaths from cancer.⁴⁴ This might explain the higher mortality rates seen at relatively young ages in the IDpop, whereas SMRs declined with increasing age, and no mortality disparity due to cancer was observed above the age of 75 years. People with ID should then perhaps be compared not with their peers of the same age from the general population but rather with older age groups to match the same degree of aging and frailty. This would then also have implications for screening recommendations for people with ID (ie, starting at younger ages). Further research is needed to test this hypothesis and to specify what would be the most appropriate comparator age groups. In particular, we need more information about the age and staging at diagnosis, which would require linking data at the individual level to cancer registry data.

A limitation of this investigation of cancer-related deaths in the Dutch IDpop is that only proximate variables instead of actual ID diagnoses could be used.⁴⁵ We used entitlements to national services that are open to people with an ID diagnosis under the assumption that the utilization of these services and underlying supportive needs are indicative for ID severity. Both demographic and mortality patterns showed substantial differences between the IDpop and the GenPop; in particular, ID-specific

causes of mortality were almost exclusively noted in the IDpop, and this confirmed accurate identification of ID cases. The subgroup with mild ID, however, under-represented older people with mild ID and people with mild ID who did not use any of the available national supportive services. Consequently, these individuals were included in the GenPop and could potentially have contributed to an underestimation of the true cancer-related mortality in the population with mild IDs.

A major strength of the current study was that we were able to analyze mortality data from the complete Dutch adult population, including an entire ID subpopulation (ie, service users). It was not necessary to rely on smaller (convenience) samples, which have been criticized before for their limited generalizability in ID research.^{4,17,46} Moreover, all mortality data were processed in a standardized way with *ICD-10* coding, and there was a relatively low amount of missing data (ie, unknown causes of death). However, the size of the ID subgroups and the length of the follow-up limited the possibilities for examining cancer-specific mortality within these subgroups. Future studies in this area also will require longer follow-up periods to investigate whether developments in cancer diagnostics and targeted cancer therapies will have similar (beneficial) effects within ID populations as they have in the general population.⁴⁷

In conclusion, cancer is the most prominent cause of death for people with ID and, in comparison with the

general population, is up to 1.5 times as likely to be the cause of death. In particular, relatively more deaths were caused by cancers targeted by the national screening program, digestive and bladder cancers, and cancers of unknown primary. Although data about the incidence of cancer among people with ID are currently inconclusive, these mortality disparities reveal a need for better tailoring of cancer screening and access to and receipt of cancer care for individuals with ID.

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CONFLICT OF INTEREST DISCLOSURES

Geraline L. Leusink is a member of the board of directors for the Maastricht University Medical Center. The other authors made no disclosures.

AUTHOR CONTRIBUTIONS

Maarten Cuypers: Conceptualization, data curation, formal analysis, investigation, methodology, project administration, resources, writing—original draft, and writing—review and editing. **Bianca W. M. Schalk:** Formal analysis, methodology, writing—original draft, and writing—review and editing. **Anne J. N. Boonman:** Investigation, writing—original draft, and writing—review and editing. **Jenneken Naaldenberg:** Conceptualization, funding acquisition, supervision, writing—original draft, and writing—review and editing. **Geraline L. Leusink:** Conceptualization, funding acquisition, supervision, writing—original draft, and writing—review and editing.

DATA AVAILABILITY

This study used nonpublic microdata from Statistics Netherlands. Under certain conditions, these microdata are accessible for statistical and scientific research (fees apply). Procedures can be found at <https://www.cbs.nl/>; for further information, email microdata@cbs.nl.

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