BMJ Oncology

Cancer incidence and diagnostic characteristics in people with intellectual disabilities in the Netherlands: a national registry-based cohort study

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To cite: Cuypers M, Naaldenberg J, Banda A, *et al*. Cancer incidence and diagnostic characteristics in people with intellectual disabilities in the Netherlands: a national registry-based cohort study. *BMJ Oncology* 2025;**4**:e000686. doi:10.1136/bmjonc-2024-000686

► Additional supplemental material is published online only. To view, please visit the journal online (https://doi.org/10.1136/bmjonc-2024-000686).

Received 27 November 2024
Accepted 23 April 2025

Linked

► https://doi.org/10.1136/bmjonc-2025-000845

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ABSTRACT

Objective People with intellectual disabilities (ID) face notable health disparities, also affecting cancer care. This study is among the first to use nationwide population and cancer registry databases to compare cancer incidence in the population with ID and the general population.

Methods and analysis A population-based cohort study enrolled all Dutch adults (18+) with indicators of ID (N=187 149) and a 1:4 random general population sample without ID (N=760 907). All cancer diagnoses from 1 January 2015 until 31 December 2020 were collected from the national cancer registry to compare incidence and diagnostic details.

Results Overall, fewer incident cancer cases were found among individuals with ID than without ID (51.0 vs 104.1/10 000 person-years; adjusted OR (adj.OR) 0.79 (0.76–0.81)), with cases occurring at younger ages and being diagnosed more often at a more advanced stage than in the general population. Key distinctions from the general population include reduced odds of skin cancer (adj.OR 0.39 (0.36–0.43)) and elevated odds of cancer of unknown primary (OR 1.60 (1.29–1.98)). The fewest cancer diagnoses occurred among those entitled to long-term ID care (adj.OR 0.63 (0.60–0.66)), with those living independently being at greater risk for cancers of digestive, respiratory and female genital organs.

Conclusion Although the overall incidence of cancer in the population with ID appears lower than in the general population, significant variations exist across ID subgroups and cancer types. These differences indicate varying exposures, lower cancer awareness and barriers to healthcare for individuals with ID. Addressing these differences requires customised strategies for public health, long-term care and oncology care.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ People with intellectual disabilities (ID) experience significant health disparities, including challenges in cancer-care access and outcomes. Existing research indicates variable cancer risks in this population, but comprehensive, population-based data are limited.

WHAT THIS STUDY ADDS

⇒ This study provides a large-scale, population-based analysis using Dutch registry data, enabling stratification within the population with ID by residency status and need for supportive care. Findings reveal distinct cancer risks among ID subgroups, with the lowest incidence in those with access to long-term residential care and higher risks for certain cancers among individuals with ID living independently.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ The findings enhance understanding of the cancer burden in the population with ID and differential patterns relative to the general population. Conclusions can inform policy and clinical practice by highlighting the importance of accessible cancer-care pathways and targeted public-health interventions for individuals with ID.

INTRODUCTION

Cancer is a significant health concern for individuals both with and without intellectual disabilities (ID), with the population with ID constituting a substantial global subgroup of up to 200 million people, equivalent to 1–3% of each country's population.[1] Defined as limitations in cognitive and adaptive functioning, ID is caused predominantly by genetic or developmental disorders emerging before adulthood.[2] Individuals with ID experience health differences due to genetic and biological vulnerabilities, as well as social and systemic barriers that limit their access to equitable healthcare.[3 4] These barriers contribute to shorter life expectancy and higher rates of avoidable mortality.[5 6] Cancer is a major contributor to such differences experienced by individuals with ID, with reports of poorer screening uptake

and receipt of fewer or less-intensive cancer treatments, despite their higher cancer-related mortality rates relative to the general population.[7–9]

Several cancer risk factors are more common among—or even unique to—the ID population. First, the genetic causes of ID can also be involved in risks for tumour development, as demonstrated by the association between Down syndrome and leukaemia. Second, reduced health literacy in the population with ID and resulting unhealthy behaviours (eg, poor nutrition, smoking and unprotected sun exposure or sex) can elevate the risk of certain lifestyle-related cancers.[10–13] Finally, differences in health and healthcare access can delay the timely diagnosis of cancer in people with ID.[14 15]

To mitigate these risks, precise cancer-incidence data for the population with ID are essential to the development of customised strategies and the optimisation of guidelines. Current, high-quality data in this area are nevertheless scarce. The existing literature reveals inconsistent findings due to methodological differences and demographic factors in the population with ID (eg, younger age and higher male-to-female ratio), which complicate comparisons between populations with and without ID.[16–18] Addressing this knowledge gap requires more and better-powered studies.[19] This study is among the first to use nationwide population and cancer registry databases to investigate all incident cancer cases in the population with ID and facilitate comparisons with the general population.

METHODS

Setting and design

National demographic databases from Statistics Netherlands, the Dutch national statistics office (CBS) were used to generate a cohort consisting of all Dutch individuals 18 years of age and older with indicators of ID (IDpop) and alive on 1 January 2015, as well as a random sample from the remaining general Dutch adult population without ID indicators (GenPop). Given that this study required prior alignment (a so-called pre-match) between CBS and databases from the Netherlands Cancer Registry (NCR) in order to reduce data handling, a 1:4 ratio was chosen for our GenPop random sample to ensure sufficient statistical power (figure 1).

The presence of ID was assumed for those individuals who were entitled to long-term care or supportive services for which any type of ID (ranging from mild to profound ID) was specified as reason for calling on the particular service. This primarily concerned the utilisation of long-term care, which is a centrally coordinated system for all ID-related care in the Netherlands. A smaller proportion of individuals with ID were identified by receipt of income benefits due to a diagnosis of mild ID only. Although none of the available databases specifies the ID aetiology, application for any of these services requires a formal diagnosis of ID. Care and support for individuals with ID younger than 18 years of age is less centrally organised, making this method less suitable for identifying children with ID. This age group was therefore excluded from this study. Individuals without indicators of ID were assumed to constitute the general population. Data on cancer diagnoses were obtained from the NCR for individuals both with and without ID. The NCR captures essentially all new cancer diagnoses at the national level, based on notification from pathological, hospital and laboratory databases. After notification, trained registrars manually retrieve relevant patient and tumour characteristics from the patient's hospital medical records.

Outcomes and variables

All unique malignant neoplasms diagnosed 1 between January 2015 and 31 December 2020 in patients from this cohort were retrieved from the NCR. Malignant neoplasms were categorised according to the International Classification of Diseases, 10th edition (ICD-10). Other variables specified the year of diagnosis (for privacy reasons the precise date of diagnosis was not included), tumour stage according to the relevant classification for the specific cancer type (ie, Tumor Node Metastasis - TNM, International Federation of Gynecology and Obstetrics - FIGO, Ann Arbor) and diagnostic parameters (eg, microscopic confirmation of diagnosis).

Within the IDpop, individuals were categorised according to dominant indicator as determined at cohort inception in 2015: (1) entitled to long-term residential ID care, (2) entitled to other not ID-specific long-term care or (3) receipt of income support. This method of identifying and classifying individuals with ID in population data is described in greater detail elsewhere.[20] The population databases of Statistics Netherlands contain the sex, date of birth and, in case of death, both the date and underlying cause of death for all individuals in the cohort.

Statistics

Demographics are presented either by frequencies and percentages or by means with SDs. Differences were tested using independent t-tests for continuous variables and, for categorical variables, χ^2 tests. We calculated the person-years (PYs) that all individuals contributed to the cohort from the start to the end of follow-up on 31 December 2020, the year of first cancer diagnosis (for which 0.5 PY was assigned) or the date of death, whichever came first. The overall incidence of cancer during follow-up (ie, unique patients with cancer) was expressed as a crude rate per 10 000 PYs, and stratified by sex and age group. The occurrence of type-specific cancer (ie, all unique diagnoses of cancer) was presented separately. Binomial logistic regression was used to obtain the odds of a cancer diagnosis for the IDpop relative to the GenPop. The models were adjusted for age and sex to account for differences in the demographic profile of the IDpop relative to the GenPop. Adjusted ORs (adj.ORs) are presented, together with 95% CIs. Subgroup analyses were conducted for separate comparisons of individuals

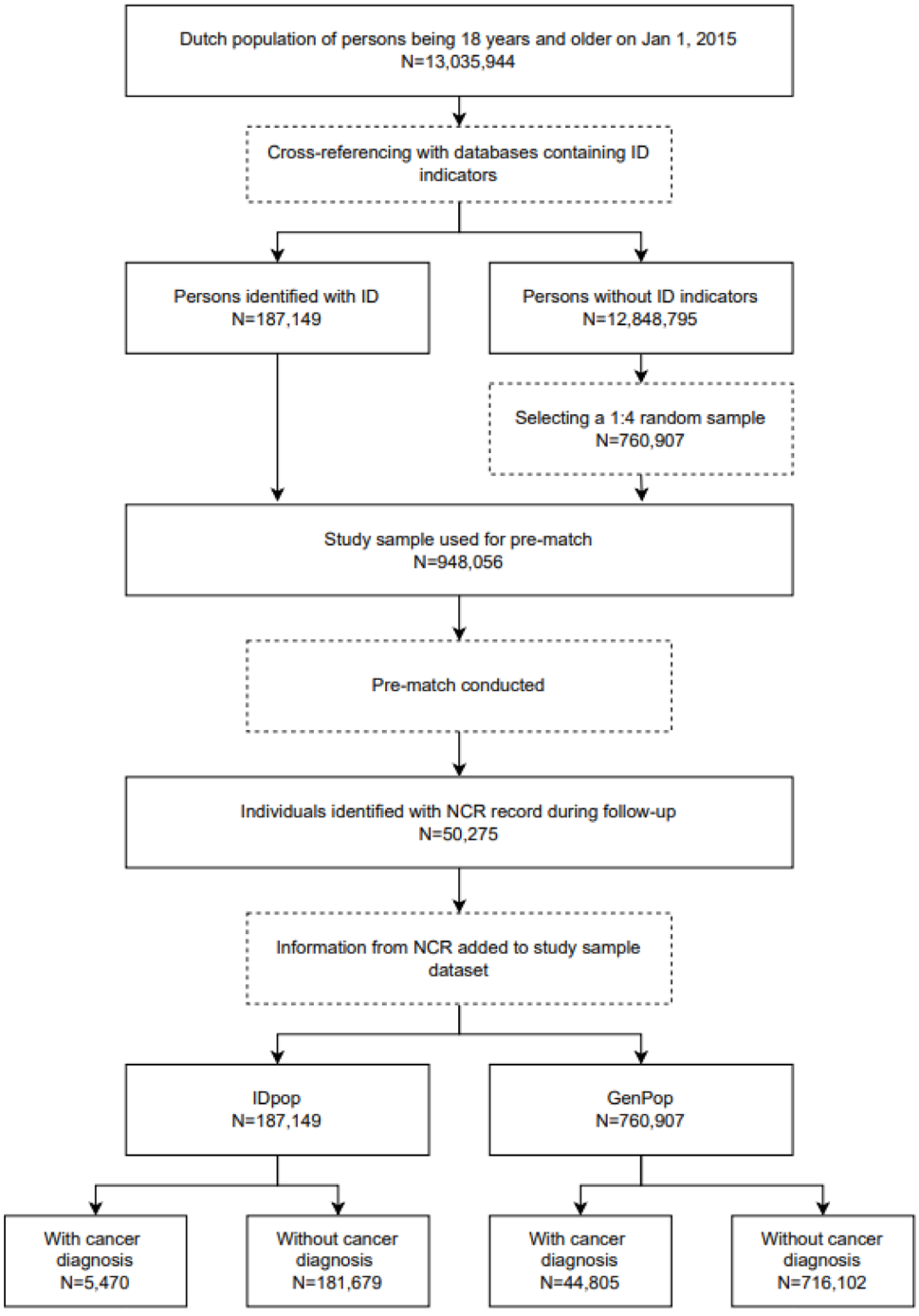

Figure 1 Flowchart of study sample composition. ID, intellectual disabilities; NCR, Netherlands Cancer Registry.

who were entitled to residential ID care at cohort inception in 2015 and those with ID who were not (ie, presumed to live independently) to the GenPop.

The distribution of all cancer diagnoses was presented by malignancy group, corresponding to the subchapters of ICD-10 Chapter II on malignant neoplasms. Three-position ICD-10 codes were used to report the most prevalent tumour sites and cancers included in the national screening programme (ie, breast, cervical and colon cancer). Variables indicating tumour stage at diagnosis were recoded and dichotomised into early stage (Stages 0, I and II) versus advanced stage (Stages III and IV). P values<0.05 were considered statistically significant. All analyses were conducted using SPSS V.25.0.

Ethics

Data collection and processing by Statistics Netherlands and NCR are exempted from obtaining individual informed consent for being public-interest tasks. The Statistics Netherlands Act governs the rights and responsibilities of Statistics Netherlands in collecting and providing access to data for research purposes. Patients have the right to view or request deletion of their data from the NCR. Our institutional ethics committee waived

Table 1 Cohort characteristics

	Individuals without ID		Individuals with ID	
N	760 907		187 149	
Sex distribution in cohort, N (%)				
Males	371 239	48.8	107 370	57.4
Females	389 668	51.2	79 779	42.6
Age at cohort start, M (SD)	48.3	17.8	39.6	15.6
Distribution across age groups, N (%)				
18–24	81 512	10.7	44 343	23.7
25–34	116 511	15.3	36 615	19.6
35–44	133 046	17.5	32 514	17.4
45–54	148 080	19.5	37 109	19.8
55–64	126 223	16.6	25 140	13.4
65–74	92 858	12.2	8770	4.7
75 year and older	62 677	8.2	2658	1.4
Primary ID indicator				
Long-term ID care with residency	0		91 064	48.7
Long-term care without residency	0		27 007	14.4
Mild ID	0		69 078	36.9

ID, intellectual disabilities.

the need for formal ethical assessment when using and combining these data sources (Reference 2017–3921), and the protocol for this study was approved by the NCR privacy review board (K18.114). We report our results in accordance with the REporting of studies Conducted using Observational Routinely-collected health Data (RECORD) Statement[21] (see online supplemental materials).

Patient and public involvement

The need for this study and its research questions arose from the recognised lack of contemporary, high-quality data and healthcare professionals' concerns about limited knowledge regarding cancer in individuals with ID. This study is embedded in a larger project on cancer and cancer screening in people with ID, for which a sounding board with multiple stakeholders is in place. Individuals with ID were not directly involved in the study's design, analysis or development of this manuscript. Co-researchers with ID will be involved in creating post-publication accessible summaries tailored to the target population.

RESULTS

In all, 187 149 individuals with ID (IDpop) and 760 907 individuals without ID (GenPop) were enrolled in this cohort. Males and individuals in younger age groups were over-represented in the IDpop relative to the GenPop (table 1). Nearly half of the IDpop had access to long-term residential ID care, and approximately one-third of the IDpop was identified through indicators of mild ID only (table 1).

During follow-up, cancer was diagnosed in 5470 individuals with ID (51.0/10 000 PYs) and in 44 805 individuals without ID (104.1/10 000 PYs), representing a statistically significant lower incidence in the IDpop (adj. OR 0.79 (0.76–0.81)). The decreased odds of cancer diagnoses were more pronounced among males with ID than among females with ID, particularly for those 45 years of age and older (table 2). Further stratification by primary ID indicators revealed that differences in cancer incidence relative to the GenPop were observed primarily among ID individuals with access to long-term residential care (adj.OR 0.63 (0.60–0.66)). In both ID subgroups without residential care, the overall cancer incidence was similar to that of the general population (adj.OR 0.94 (0.88–1.00), respectively, adj.OR 0.96 (0.92–1.00)) and non-significantly higher when excluding skin cancer (table 2).

Cancer diagnoses among people with ID were less frequently made at an early stage than in the GenPop (45.0 vs 52.1%, p<0.0001), and information on tumour stage was missing more often for individuals with ID (15.4% vs 10.7% of all cases; table 2). In addition, fewer diagnoses in the IDpop were microscopically confirmed, and a larger proportion of these confirmed diagnoses were histology confirmed metastases (table 2).

On average, individuals with ID were almost 10 years younger when diagnosed with cancer than individuals without ID (58.3 vs 67.9 years of age; table 2). Types of cancer diagnosed were distributed differently in the IDpop than in the GenPop (table 3). Melanoma and other skin cancers were the most common cluster in the GenPop, but

Table 2 Demographic and diagnostic characteristics of all patients diagnosed with cancer during follow-up

	Individuals without ID		Individuals with ID		
N	760 907		187 149		
Person-years*	4 302 826.5		1 072 122.1		Adjusted OR (95% CI)
Person-years, M (SD)	5.7	1.1	5.7	1.0	
Individuals with new cancer diagnosis, N (%)	44 805	5.9	5470	2.9	0.79 (0.76 to 0.81)
Cancer incidence per 10 000 PYs, OR	104.1		51.0		
Sex, N (%)					
Males	22 316	6.0	2964	2.8	0.78 (0.75 to 0.81)
Females	22 489	5.8	2506	3.1	0.82 (0.79 to 0.86)
Age at diagnosis, M (SD)	67.9	13.0	58.3	12.7	<0.0001
Cancer diagnoses per age groups†, N (%)					
18–24	338	0.4	176	0.4	0.98 (0.81 to 1.17)
25–34	1010	0.9	330	0.9	1.08 (0.96 to 1.23)
35–44	2782	2.1	669	2.1	1.03 (0.95 to 1.12)
45–54	6696	4.5	1515	4.1	0.92 (0.87 to 0.98)
55–64	11 617	9.2	1814	7.2	0.76 (0.72 to 0.80)
65–74	13 795	14.9	795	9.1	0.55 (0.51 to 0.60)
75 year and older	8567	13.7	171	6.4	0.42 (0.36 to 0.49)
Primary ID indicator (GenPop is reference group), N (%)					
Long-term ID care with residency	0		2311	2.5‡	0.63 (0.60 to 0.66)
Long-term ID care without residency	0		950	3.5	0.94 (0.88 to 1.00)
Mild ID (income support only)	0		2209	3.2	0.96 (0.92 to 1.00)
Cancer stage at diagnosis, N (%)§					
Early-stage	25 809	52.1	2630	45.0	<0.0001
Advanced stage	18 404	37.2	2317	39.6	
No information available	5325	10.7	900	15.4	
Microscopic confirmation (ie, biopsy available), N (%)					
Microscopic confirmed	46 648	90.2	5288	85.6	<0.0001
Non-microscopic confirmed	2890	5.8	559	9.6	
Histological confirmation of metastases only	1976	4.0	280	4.8	

*Person-years calculated until first cancer diagnosis, death or end of follow-up.
†Age groups refer to age at enrolment, see table 1.
‡Percentage refers to size of subgroup, see table 1.
§Based on available clinical or pathological stage in relevant classification (ie, TNM, FIGO, or Ann Arbour) and dichotomised (Stage 0–I–II vs Stage III–IV). Based on unique cancer diagnoses, multiple diagnoses per individual possible during follow-up adj.ORs when excluding skin cancer 0.71 (0.67–0.74)/1.08 (1.01–1.16)/1.07 (1.02–1.12).
Adj.OR, adjusted OR; FIGO, International Federation of Gynecology and Obstetrics; GenPop, general population; ID, intellectual disabilities; PYs, person-years; TNM, Tumor Node Metastasis.

the fourth most common in the IDpop. Cancer of digestive and respiratory organs was the most common cluster in people with ID. As for specific types of cancer, the IDpop exhibited elevated risk for cancer of unknown primary (CUP; adj.OR 1.60 (1.29–1.98)) and oesophageal cancer (adj.OR 1.26 (1.06–1.49)), as compared with the GenPop. When stratifying the ID population by living situation (ie, residential care and independent living), individuals in residential care had similar or lower risks of the most common cancer types, except for CUP, relative to the GenPop. In contrast, individuals with ID living independently exhibited a higher risk of lung, pancreatic, oesophageal cancer and CUP (figure 2). The risk of cervical cancer was non-significantly higher (figure 2), with a significantly higher risk for the cluster of all female genital cancers (C51-C58) (OR 1.33 (1.13–1.56); (see online supplemental table 1). All outcomes of this stratified comparison are presented in online supplemental table 1.

DISCUSSION

This study combines Dutch national demographic data with cancer registry data to estimate the incidence of

Table 3 Distribution of cancer diagnoses by ICD-10 subchapter and most common cancer types

	Individuals without ID		Individuals with ID		
N	760 907		187 149		
Total amount of cancer diagnoses*	48 751		5772		Adjusted OR (95% CI)
Distribution of all cancer diagnoses by ICD-10 subchapter, N (% of total number of diagnoses)					
C00-C14 - lip, oral cavity and pharynx	883	1.8	138	2.4	0.96 (0.79 to 1.15)
C15-C26 - digestive organs	8643	17.7	1304	22.6	1.04 (0.98 to 1.11)
C30-C39 - respiratory and intrathoracic	5417	11.1	798	13.8	1.01 (0.93 to 1.09)
C40-C41 - bone and articular cartilage	49	0.1	14	0.2	1.21 (0.66 to 2.22)
C43-C44 - melanoma and other malignant skin	11 425	23.4	603	10.4	0.39 (0.36 to 0.43)
C45-C49 - mesothelial and soft tissue	244	0.5	52	0.9	1.28 (0.94 to 1.74)
C50-C50 - breast	6205	12.7	766	13.3	0.78 (0.73 to 0.85)
C51-C58 - female genital	1882	3.9	337	5.8	1.18 (1.05 to 1.33)
C60-C63 - male genital	4555	9.3	474	8.2	0.60 (0.55 to 0.66)
C64-C68 - urinary tract	3792	7.8	423	7.3	0.78 (0.70 to 0.86)
C69-C72 - eye, brain and other CNS	1366	2.8	248	4.3	0.97 (0.84 to 1.12)
C73-C75 - thyroid and other endocrine	387	0.8	70	1.2	0.83 (0.64 to 1.07)
C76-C80 - ill-defined and unspecified	556	1.1	111	1.9	1.62 (1.31 to 2.00)
C81-C96 - lymphoid, and haematopoietic	2709	5.6	345	6.0	0.77 (0.69 to 0.87)
C97-C97 - independent multiple sites (C97)†	–		–		
D00-D09 - in situ neoplasms†	–		–		
D10-D36 - benign neoplasms†	–		–		
D37-D48 - uncertain of unknown behaviour	638	1.3	89	1.5	0.96 (0.77 to 1.21)
Most common tumour sites and screening cancers					
C44 - skin	11 425	23.4	603	10.4	0.39 (0.36 to 0.43)
C50 - breast (female only)	6205	12.7	766	13.3	0.78 (0.72 to 0.84)
C34 - lung	4849	9.9	712	12.3	1.02 (0.94 to 1.11)
C61 - prostate	4236	8.7	328	5.7	0.49 (0.44 to 0.55)
C18 - colon	3498	7.2	480	8.3	0.98 (0.89 to 1.08)
C67 - bladder	2388	4.9	254	4.4	0.76 (0.67 to 0.87)
C20 - rectum	1353	2.8	199	3.4	0.94 (0.81 to 1.10)
C64 - kidney	1006	2.1	137	2.4	0.83 (0.69 to 1.00)
C25 - pancreas	987	2.0	138	2.4	1.04 (0.87 to 1.25)
C83 - lymphoma	953	2.0	90	1.6	0.59 (0.47 to 0.74)
C15 - oesophagus	887	1.8	169	2.9	1.26 (1.06 to 1.49)
C53 - cervix uteri	304	0.6	58	1.0	0.85 (0.64 to 1.14)
C80 - unknown primary	551	1.1	108	1.9	1.60 (1.29 to 1.98)

All analyses with sex-specific cancer types are carried out in the relevant sex group and adjusted for age only.
Males with C50 n=53.
*Patients could contribute multiple cancers during follow-up.
†Diagnoses in these categories are not captured by the Netherlands Cancer Registry.
CNS, Central nervous system; ICD-10, International Classification of Diseases 10th edition; ID, intellectual disabilities.

cancer among people with ID and to make comparisons with the general population. Results indicate a lower overall incidence of cancer among people with ID, with variations in age, cancer type and residential-care status (serving as a proxy for ID severity).

To date, the available scientific literature has been inconclusive on the incidence of cancer among people with ID. Although results come from different countries and care settings, and although they span several decades, outcome discrepancies are largely attributable to differences in the definition and selection of populations with ID and to variations in study characteristics (eg, sample size and data sources).[16–18] A study with a relatively small group of individuals with

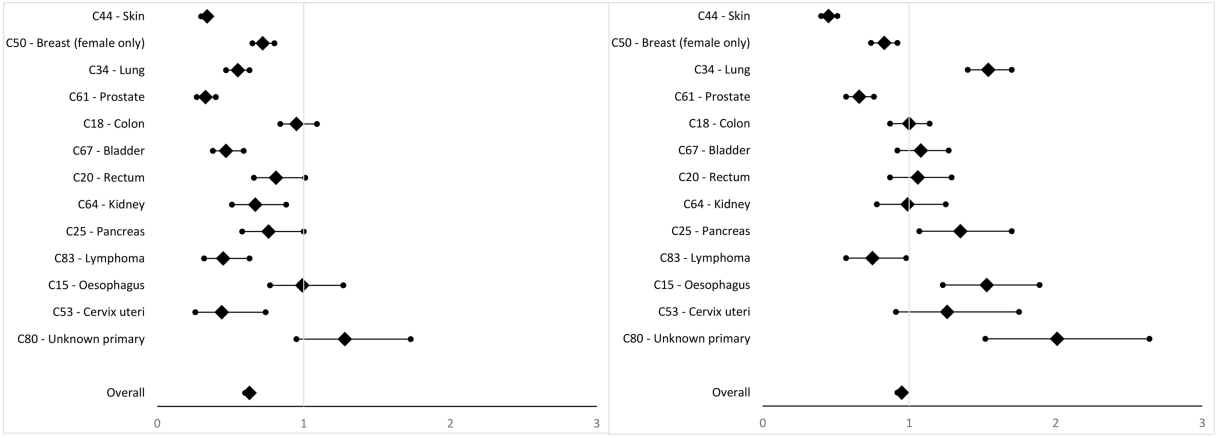

Figure 2 Adjusted ORs for most common cancer sites for residential care ID-group versus general population (left) and non-residential care ID-group versus general population (right). ID, intellectual disabilities.

ID or a low number of incident cases may either overestimate or underestimate true differences, particularly when compared with a large control group of individuals without ID. Our results identify a relatively high number of individuals with ID, and the aforementioned biases are addressed with analyses adjusted to account for demographic differences between these groups. We observed that cancer incidence peaked approximately 10 years earlier in the ID population than in the general population. Cancer incidence in the population with ID was highest between the ages of 55 and 64 years, as compared with 65 and 74 years in the general population. Overall, cancer risks declined for people with ID relative to the general population with increasing age, with onset from 45 years of age onwards. Given the lower life expectancy of individuals with ID (estimated at 15 years shorter than the general population), individuals with ID who reach older ages are potentially more likely to be in relatively good health, thus potentially indicating a healthy-survivor effect in the cohort with ID providing an explanation for the growing difference in cancer incidence at older ages between people with and without ID.[22]

Whereas we found a lower overall cancer incidence in people with ID, a previous study in the same population and recent findings in Scotland indicated a high risk of cancer-related death compared with the general population.[7 23] This apparent discrepancy might be due to differences in the types of cancer diagnosed. In line with the findings in Scotland, we observed a much lower incidence of skin cancer (which typically has a low mortality rate), but a relatively higher incidence of cancers with high mortality rates, including CUP and gastrointestinal cancers.[23–25] Although fewer people with ID were diagnosed with cancer overall, the types of cancers diagnosed carry higher mortality risks.

In addition to differential risks of developing cancer in people with ID, our results point to potential risks of diagnostic delays and fewer diagnostic procedures. Among people with ID, fewer cancer diagnoses were made at an early stage and information about tumour stage was more often missing. These findings are in line with recent findings reported elsewhere.[15 26 27] Missing stage data, but also observing fewer microscopically confirmed cancer cases might reflect more than missing data alone, but could indicate incomplete diagnostics or staging procedures. At the individual level this can impact treatment options, and at the population level it contributes to incomplete understanding of the true cancer burden among people with ID as it limits accurate comparisons with the general population. With regard to the timing of diagnosis, people with ID were at increased risk for being diagnosed with CUP. This type of cancer represents confirmed, metastatic cancer for which the primary tumour site cannot be identified and it typically has a poor prognosis.[28] Missed early symptoms and late presentation at an oncology provider are common aspects of CUP.[29] The elevated risk of CUP was found in all ID subgroups, but it was particularly high for people with ID without access to residential ID care and who were thus living independently with less structured daily support. This subgroup appears to encounter challenges in timely signalling of symptoms, promptly seeking medical assistance and navigating the healthcare system.[30 31]

In addition to a higher incidence of CUP, the subgroup of individuals with ID living independently also exhibited elevated incidences of lung, pancreatic and oesophageal cancers, relative to the general population. In contrast, the incidence of all cancer types was lower or similar to that of the general population in those with access to residential ID care. Lifestyle factors likely contribute to these differences.[10 32] The higher incidence of lung cancer among people with ID living independently might be associated with higher smoking rates in this subgroup, while pancreatic and oesophageal cancers could be linked to unhealthy diet and increased alcohol consumption.[11 33 34] However, due to the population-based design of this study, we lacked data on the prevalence of these lifestyle factors within our sample. Accessing such information would require linkage with health records (eg, general practitioner or hospital data). As a result, we were unable to assess their potential confounding effects.

Current findings also have implications for national screening programmes. The relevance of participation in cancer screening by people with ID could be a topic of debate, particularly if it is assumed that their overall risk of developing cancer is lower. Several international studies have shown poor participation rates in cancer screening among this population, with contributing factors including physical barriers, lack of knowledge and insufficient customisation of information and procedures to meet their needs.[9 35] In many countries, including the Netherlands, population screening programmes target colon, breast and cervical cancer. Our findings indicate differential outcomes for each of these cancer types. Risks for colon cancer appeared equal between people with and without ID, regardless of ID subgroup. In contrast, the risk of breast cancer was lower, particularly among those in residential ID care, as compared with those living independently. Although the risk of cervical cancer was not significantly lower in the population with ID, the incidence was significantly lower in the subgroup in residential ID care, but higher (non-significantly) among others with ID. This suggests that individual counselling in residential care settings could be beneficial, while people with ID living independently should be encouraged to participate in all screening programmes, but may require customised information provision and screening methods.[36] Beyond population screening, there is a public health task to raise awareness of skin cancer among people with ID and those around them. The incidence of skin cancer was substantially lower in this group, even though people with ID are potentially at heightened risk due to suspected unhealthy behaviours, including unprotected exposure to sunlight and insufficient awareness of suspicious skin spots.[13]

The main strength of this study was the combination of multiple population data sets, making it one of the few studies in the field of cancer and ID to combine multiple national data sets.

Limitations are related to the identification of individuals with ID. Like most other countries, the Netherlands has no national register of people with disabilities. We therefore had to derive the presence of ID based on entitlements to ID-specific supportive care and receipt of income support, although without information about ID aetiology. If this information were to be available, it could provide more information on the causal pathways from ID aetiology to tumour development, in addition to guiding the development of customised policies. A recent review highlights the relevance of investigating these relationships in greater detail.[37]

Our identification method also constrained our ability to include more individuals with mild ID and those who did not use support from national systems, like many children with ID. This implies our results may not fully represent the entire spectrum of individuals with ID, and underestimate cancer risks in specific subgroups. Finally, the length of follow-up in this study was limited to the availability of data containing ID indicators (from 2015 onwards) and completed years in the cancer registry (up to 2021 at the time of data collection). Moreover, only the year of diagnosis was available meaning a precise time-to-event could not be calculated. Including this information, having a longer follow-up, and availability of information on other risk factors (eg, comorbidities, life style factors) could allow future investigation of lifetime risks, causal relations and the detection of changes over time.

CONCLUSION

Although the overall cancer incidence in the population with ID is apparently lower than in the general population, significant variations emerged in specific ID subgroups and for certain cancer types. This suggests differences in exposure, reduced awareness of cancer risks and obstacles to healthcare and early cancer diagnostics for individuals with ID. Addressing these differences requires customised strategies spanning public health to oncology care.

Contributors MC, JN, HB and GL conceptualised and designed the study. MC, AB and LO had full access to the data and contributed to the investigation. MC performed formal analyses. JN, HB and GL provided supervision. All authors participated in writing, reviewing and revising the manuscript and approved the final version. MC is the guarantor.

Funding This study was funded by grants from the Dutch Ministry of Health, Welfare and Sport (VWS; Grants 329574 and 90000803) and the Netherlands Organisation for Health Research and Development (ZonMw; Grant 641001100).

Disclaimer Funding organisations had no role in the study design, data collection, data analysis, data interpretation or writing of this report.

Competing interests None declared.

Patient and public involvement Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

Patient consent for publication Not applicable.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement Data may be obtained from a third party and are not publicly available. Data were retrieved from the Netherlands Cancer Registry (NCR) and linked to population data from Statistics Netherlands (CBS) for the specific purpose of this study. Results are based on calculations by Radboud University Medical Center in project number 8359 using non-public microdata from Statistics Netherlands. Original datasets contain records that have been pseudonymised at the individual level and stored in a secure digital research environment at Statistics Netherlands. Under certain conditions, these microdata are accessible for statistical and scientific research. The same original data can be requested from the NCR and CBS according to standard procedures (fees apply), see for further information: https://www.cbs.nl/en-gb/our-services/customised-services-microdata/microdata-conducting-your-own-research.

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