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The importance of cancer prevention policies to inform and guide preventative and screening measures for people with intellectual disabilities: The COST project “Cancer- Understanding Prevention in Intellectual Disabilities”

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Abstract

Cancer is a global public health problem, but its exact prevalence in people with intellectual disabilities is still uncertain. This population, with limited health skills and complex health needs, faces many challenges in cancer prevention, screening, timely diagnosis and treatment. Furthermore, they are often underrepresented in general cancer prevention and screening policies across Europe, leading to widened disparities in health outcomes and premature mortality. Thus, unified national and local policies are needed to reduce inequalities and promoting a pan-European inclusion of people with intellectual disabilities. Our goal is to raise public awareness of this issue, including the involvement of people with intellectual disabilities, and promote engagement from relevant stakeholders. The COST Action 'Cancer- Understanding Prevention in Intellectual Disabilities' (CUPID) project will address health inequalities faced by people with intellectual disabilities in relation to cancer, and support the development of policy recommendations specifically tailored to their unique cognitive and healthcare needs, having a positive long-term impact on quality of life.

Keywords

cancer, intellectual disability, policy, prevention, screening

Introduction

Cancer is a major public health problem, responsible for over 1.3 million deaths in the European Union (EU) every year, making it the second leading cause of death ([ECIS – European Cancer Information System, 2020](#)). Many EU countries identify cancer prevention and screening as a key public health strategy to address this problem ([European Commission, 2021](#)). Populations with limited health literacy and more complex health needs, such as people with intellectual disabilities, are potentially underserved by these generic strategies. This may contribute to health inequalities in terms of preventative care, timely diagnoses, and adequate treatment, leading to a widening of disparities in health outcomes and premature mortality ([Krahn and Fox, 2014](#); [Heslop, Byrne, et al., 2022a](#)). Specifically, a significant proportion of cancer-related deaths could be avoided in this population if timely and adequate access to prevention and treatment services was provided ([Cuypers et al., 2022](#)). In the general population, cancer stage at diagnosis (and the initiation of treatment) has a significant impact on 5-year survival rates, although the influence of the stage at diagnosis on outcomes does vary by cancer type ([Nuffield Trust, 2023](#)). People with intellectual disabilities are usually diagnosed with cancer at a more advanced stage than the general population ([Satgé et al., 2014](#); [Willis, Samalin and Satgé, 2018](#)).

Having an intellectual disability is characterised by significant limitations in both intellectual functioning and adaptive behaviour in conceptual, social and practical skills. This disability is functionally due to the developmental period defined before the individual reaches adulthood ([Schalock, Luckasson and Tassé Marc, 2021](#)). In upper-middle and high income countries, an intellectual disability is estimated to occur in every 15 out of 1,000 births ([McKenzie et al., 2016](#)). The European Commission's Strategy for the Rights of Persons with Disabilities 2021-2030 among other things, underlined the necessity to remove barriers to healthcare access for people with intellectual and other disabilities to ensure equal treatment with the broader population ([European Commission Directorate-General for Employment Social Affairs and Inclusion, 2021](#)). Despite this call for action, and the efforts of the European Union and its Member States, people with intellectual

disabilities continue to face widespread barriers in accessing health and related services, such as those for health care (including medical care, therapy, and assistive technologies). These barriers may originate from: insufficient legislation, strategies and policies; inadequate service provision; difficulties with delivery of services; ignorance about disabilities; discrimination and negative attitudes; inaccessibility; lack of funding; and insufficient participation in decisions directly affecting their lives (World Health Organization, 2015). It is still unclear to what extent existing health policies adequately address the specific need(s) of people with intellectual disabilities. Therefore, work is urgently needed to improve these services across Europe and shape them to the specific health and other needs of people with intellectual disabilities so that cancer care is equitable for this vulnerable population. Our view is that this aim would be best achieved through collaborative work involving researchers, healthcare professionals, service providers, educators, policy makers, interdisciplinary stakeholders, people with intellectual disabilities and their representatives. This is an important public health issue that people with intellectual disabilities are facing in terms of cancer prevention, screening, timely diagnosis and treatment. There is therefore a need to focus on research into the diverse need(s) of people with intellectual disabilities in regard to cancer prevention and screening.

Relevance of cancer prevention policies

According to current estimates, around 40% of all cancers can be prevented (ECIS – European Cancer Information System, 2020). Cancer prevention is the most effective and long-term strategy for cancer control. Using individual-level interventions may provide complementary benefits to the mass benefits of the population-level interventions, but evidence shows that asking all eligible people to get screened has an impact on mortality (Armaroli et al., 2015). Therefore, a comprehensive cancer control addresses the entire population while trying to meet the needs of different at-risk subgroups. Four levels of interventions to deal with inequalities were described by Whitehead, including activities to strengthen individuals, communities (including the health of the disadvantaged communities), improving conditions of living and work environments across the whole population, and promoting healthy macro-policy. Policies can address issues at all these levels, by allocating resources to evidence-based interventions and policy instruments (Whitehead, 2007; de Leeuw, Clavier and Breton, 2014; Hillier-Brown et al., 2014). The most important components of a cancer prevention strategy are national policies and programmes to reduce exposure to cancer risk factors, raise awareness by providing people with information and support their need to adopt healthy lifestyles, and implement governance and decision-making processes in cancer screening as fundamental pillars of an effective cancer prevention strategy (World Health Organization, 2023a). Cancer screening governance is recognized as a critical tool to enhance success in combating cancer and reducing health inequalities. Therefore, establishing and supporting more structured and well-defined policy-making and governance structures are essential prerequisites for the sustainability and efficacy of cancer screening programs (Lönnerberg et al., 2017).

Whilst a universally accepted definition of the nature and character of policy is lacking (de Leeuw, Clavier and Breton, 2014), it may be broadly agreed that policies which pertain to health aim “to improve the conditions under which people live: secure, safe, adequate, and sustainable livelihoods, lifestyles, and environments including housing, education, nutrition, information exchange, child care, transportation and necessary community and personal social and health services” (Milio, 2001). Policies provide a course of action which can guide and influence decisions,

as policies are derived from standards of practice, law and regulations (O'Donnell and Vogenberg, 2012). For policies to be effective, there needs to be a clear understanding of how and why they have been developed as well as being easily implemented and complied with (O'Donnell and Vogenberg, 2012). The World Health Organisation (WHO) recommends that, to control cancer, there is a need to have national policies, plans and programmes (World Health Organization, 2023b) through a National Cancer Control Programme (NCCP); however substantial international heterogeneity exists in the extent that cancer control structures have been implemented in the EU, and there is no widely agreed format for a NCCP (Espina et al., 2018).

As the European Commission's Strategy for the Rights of Persons with Disabilities 2021-2030 (European Commission Directorate-General for Employment Social Affairs and Inclusion, 2021) makes clear, appropriate access to cancer preventative and screening activities should be available to everyone. Even though some EU member states have implemented population-based national screening programmes for prevalent cancers (namely breast, cervical and colorectal cancer), inequalities in screening uptake are substantial, both between countries and within countries across population groups (Berchet et al., 2023). The existing data are often variable or incomparable across different healthcare (and social) systems and countries. In this context, Ponti et al. identify a significant inequality across Europe when it comes to access to prevention activities for people with intellectual disabilities (Ponti et al., 2017). Also, comparative research between Norway, Romania, England and Scotland concludes that the availability and quality of care for people with intellectual disabilities is substantially different between these countries (Brown et al., 2017). In addition, adequate governance, legal frameworks and policy-making structures, as well as quality data on cancer prevention and screening programmes are limited or mostly lacking for many countries across Europe (Lönnberg et al., 2017), particularly middle-income and EU candidate countries.

Challenges for population with intellectual disabilities

The exact prevalence of cancer in people with intellectual disabilities is uncertain, yet several studies identify cancer to be among the leading causes of death in this population (Trollor et al., 2017; Cuyper et al., 2022; McMahan et al., 2023). There are many reasons for the high cancer mortality rate among people with intellectual disabilities, for example, cancer diagnoses in this population are often made at a more advanced disease stage compared to the general population, and cancers of unknown primary source are more prevalent, suggesting that early diagnosis is more difficult to reach for people with intellectual disabilities (Liu et al., 2021).

However, earlier cancer diagnosis for people with intellectual disabilities can improve their health outcomes. When detected in early stages, non-metastasized cancers often have surgical, radiotherapeutic, immuno- or combination therapies available with curative intent. If progressed to more advanced stages, in particular with metastases at distance from the primary tumor location, treatment usually is of palliative nature, intended to minimise symptom burden. Particularly, primary healthcare-based systems are associated with controlling costs and improving health outcomes, especially for disadvantaged populations. Many high-income countries have adopted a strategy of focusing more on primary healthcare to reduce health inequalities. Well-supported primary healthcare for people with intellectual disabilities can prevent unnecessary hospitalizations. For instance, in Canada, the likelihood of hospitalization for 'ambulatory care-sensitive conditions' in people with intellectual disabilities is found to be significantly reduced with effective primary care that should 'delay progression or prevent serious complications.' Therefore, improving access and the uptake of preventive services like cancer screening through

effective primary healthcare delivery can contribute to early diagnosis and better health outcomes for individuals with intellectual disabilities (Lennox, Van Driel and van Dooren, 2015). This was illustrated by a population-based study by Heslop et al. who reported that in a cohort of deceased adults with intellectual disabilities from England, most cancer diagnoses were made at a late stage, frequently as a result of an emergency presentation, meaning fewer treatment options were available. This is problematic especially as there is a national screening programme aiming to detect colon cancer early (Heslop, Cook, et al., 2022b). Improving and enhancing the access to early detection cancer screening for people with intellectual disabilities might improve the treatment possibilities and outcomes. Additionally, people with intellectual disabilities have lower cancer screening rates compared to the general population, all over the world (Steele et al., 2017; Iezzoni et al., 2021). Particularly, it has been reported that the participation rates in breast and cervical cancer screenings are significantly lower in women with severe intellectual disabilities compared to the general population, but there is no significant difference between the screening participation rates between the general population and individuals with mild intellectual disabilities (Maltais, Morin and Tassé, 2020).

There also appears to be a relationship between certain (genetic) intellectual disability syndromes (e.g. Down's syndrome, Neurofibromatosis type 1) and risk of tumour development, including particular high incidence rates of leukaemia, melanoma, and colorectal cancers in certain subgroups of people with intellectual disabilities (Hasle et al., 2016; Landry et al., 2021). Moreover, an increased risk of cancer among people with autism spectrum disorder is suggested, most likely due to the pleiotropic effects of autism-associated genes, i.e., overlap of genes underlying autism and genes that affect cancer risk (Liu et al., 2022).

Although cancer screening services are widely available and valued in many countries, inequalities in cancer screening and access to cancer screening have been reported between people with and without intellectual disabilities (Chan et al., 2022). The WHO reported four major causes of health inequalities faced by individuals with intellectual disabilities: 1) structural factors, including laws and policies, 2) social determinants of health, involving gaps in formal social support mechanisms, 3) risk factors for non-communicable diseases in particular, and 4) barriers in accessing healthcare (World Health Organization, 2022). Additionally, previous research on specific barriers to cancer screening in the context of disability, identified the following factors: individual (e.g. anticipatory anxiety regarding the procedure, inability to perform one's own breast self-examination or negative attitudes towards screening), interpersonal (e.g. inadequate patient-provider communication and training to know the needs of people with intellectual disabilities), and environmental barriers (e.g. lack of reliable transportation to screening facilities and inaccessible medical equipment) contributing to the health inequalities (Merten et al., 2015; Chan et al., 2022). Participation in national cancer screening programmes is therefore markedly lower than in the general population (Horsbøl et al., 2023).

High health care costs are often incurred in relation to chronic disease; however, costs for adults with disabilities appear disproportionately large compared to the wider population, raising questions as to the cause of this differential. In this context, Anderson et al. (Anderson et al., 2010) note that using preventive care services and health promotion interventions, accompanied by improving access to medical care for people with disabilities can reduce the incidence of secondary conditions that cause disability through early diagnosis and intervention thereby implicitly reducing costs overall.

In general, people with intellectual disabilities need support in performing social, communication and interpersonal activities, as well as activities of daily living (Chan et al., 2022). It has been reported that people with intellectual disabilities face difficulties in benefiting from all health

services, including cancer screening. For example, it is reported that people with intellectual disabilities who are more dependent in performing activities of daily living are less likely to undergo mammography (Wilkinson et al., 2011). People with intellectual disabilities often require support to access healthcare and their caregivers/families are often untrained in cancer prevention and screening procedures. Due to poor health literacy people with intellectual disabilities are not aware of, and often cannot express their cancer symptoms (Cuypers et al., 2020). Globally, healthcare services need to support clinicians who are involved in the care of people with intellectual disabilities, on how to communicate effectively, to educate and support people with intellectual disabilities, especially in cancer prevention. If the evident lack of training and education in the field of intellectual disability is not revised, this may result in poor health outcomes for this group (Howie et al., 2021).

Inherent in the European Commission's Beating Cancer Plan (2021) (European Commission, 2021) is a political commitment to turn the tide against the impact of cancer on the population and reduce inequalities in healthcare access and treatment. Although people with disabilities are referenced in the main text of this plan, it may be argued that it does not effectively respond to the broader pre-existing structural and attitudinal inequalities that exist in terms of prevention, early detection, diagnosis and treatment that people with intellectual disabilities may experience (European Disability Forum, 2022).

Overall, there remains a general absence in the policy landscape across Europe specifically concerning people with intellectual disabilities. This absence in policy may arise from a lack of contemporary data surrounding the optimal cancer care and decision making in this population (Boonman et al., 2022; Heslop, Cook, et al., 2022b). In parallel, most European countries have laws prohibiting discrimination on the basis of disability and all European countries have signed and ratified the UNCRPD protocol (2006) (United Nations, 2006) which sets out under Article 25a that people with disabilities should be provided with "...the same range, quality and standard of free or affordable health care...", but suboptimal cancer care is still a serious problem (Reeves and Collingridge, 2022). The disconnect between being included in policy and actual transformation is wide and there is a critical need for people with intellectual disabilities to be heard and placed centrally within policy at an EU, national and local level.

Equitable healthcare requires comprehensive understanding of the epidemiology of health conditions in the intellectual disability population (United Nations, 2006). Research into the aetiology and epidemiology of cancer in people with intellectual disabilities is not well understood compared to the general population and, as a result, has clear implications in terms of prevention and treatment. Significant gaps in knowledge and methodological limitations for many health conditions as well as across different countries indicate that the population with intellectual disabilities is underrepresented in most cancer prevention policies (Liao et al., 2021). Low levels of awareness of the disease and its risk factors may expose people with intellectual disabilities to underdiagnosis, misdiagnosis, inappropriate pharmaceutical interventions and missed opportunities for preventive healthcare.

The COST 'Cancer- Understanding Prevention in Intellectual Disabilities' (CUPID) Action

To overcome the evident health inequalities faced by people with intellectual disabilities in relation to cancer, the COST CUPID Action project has been established in 2022 (Wells, 2022). CUPID, funded by COST Actions, and brings together expertise from across Europe and the wider world.

The underrepresentation of people with intellectual disabilities in cancer prevention policies

The aim of the COST CUPID Action is to generate systematic knowledge on the extent to which cancer prevention strategies across Europe need to be specifically tailored to the unique cognitive and healthcare needs of people with intellectual disabilities, and their right to equitable healthcare (Wells, 2022). It is not contested that people with intellectual disabilities are underrepresented in most cancer prevention policies. This is particularly important as the epidemiological evidence appears to indicate that people with intellectual disabilities are at a higher risk of developing specific cancers (Liu et al., 2021). There is also an increased recognition of potentially poorer cancer outcomes in people with intellectual disabilities (Heslop, Cook, et al., 2022b). People with intellectual disabilities are now living longer, which may lead to an increase in cancer incidence as it is predominantly a disease of older age. From this perspective, if Europe's Beating Cancer Plan is to be truly transformative and inclusive as opposed to tokenistic for people with disabilities, the COST CUPID Action consortium aims to operationalise calls from the European Disability Forum (2022, p.3) (European Disability Forum, 2022) to:

- collect disaggregated data and statistics on cancer screening and care;
- adopt measures to promote and support disability-inclusive health services, including cancer prevention, screening and care;
- introduce accessible public health campaigns and communication of cancer measures;
- promote and support the training of healthcare professionals and people working with persons with disabilities; and
- consult representative organizations of persons with disabilities.

Recognising the growing challenge that cancer in this population of people with disabilities presents, requires resilient, accessible, effective and evidence-informed policies. There is a critical need for this change to be driven by the European Union for universal action and dissemination to ensure that EU, national and local policies are aligned to reduce inequalities and inequities and promote a pan European inclusion of people with intellectual disabilities. This should be underpinned by a proportionate universalism philosophy (Marmot, 2010) whereby the resourcing and delivery of policy actions are at a scale and intensity appropriate to the needs of people with intellectual disabilities.

The CUPID's aim will be achieved through several approaches intended to ensure that its activities have sustained and meaningful impact. These will include an acknowledgment of the effects of social determinants of health on the lives of people with intellectual disabilities, as well as recognizing the value of activity co-design. As such, the project will bring together people with intellectual disabilities and carers to work with other stakeholders, to ensure that 'experts by experience' play a key role in the project.

Working group 2 aims

Working Group 2 (WG2) is one of four working groups within the CUPID and will focus on cancer prevention policies across Europe. The aim of WG2 is to identify and report on the variation in policies and services across countries and assess the extent to which people with intellectual disabilities can access cancer prevention services equitably, which is anticipated to vary substantially between nations. It will also report on any adjustments to service design and delivery that

are made to ensure prevention initiatives can be accessed by people with intellectual disabilities. The WG2 will move on to identify areas in need of change, including any necessary training and additional research.

Data capturing

The initial work of CUPID WG2 will be a scoping exercise, collating information on relevant policies on cancer prevention across Europe. This activity will map all European countries on their general health system, any population-wide policies and programmes aimed at cancer prevention and screening, and any national action related to cancer and people with intellectual disabilities, including law, policies, strategies or guidance. Subsequent WG2 activities will comprise a systematic collation of quantitative and qualitative research evidence on cancer prevention and screening for people with intellectual disabilities in Europe, including the specific challenges they face, and any service interventions or adjustments that have been used in response to these challenges and needs. Multiple digital scientific databases will be searched, using a structured search strategy comprising MeSH and free-text terms related to key elements of the search (e.g. intellectual disabilities; cancer prevention or screening; barriers and facilitators), as well as Grey Literature sources, such as Open Grey and Google Scholar. Explicit entry criteria will be used to reduce the potential for indexing and publication biases. The value of the completed work will be not only in having an overview of the current European situation, including any international variation, but also in the sharing of ideas for policy adjustment and/or further research; both aspects are intended to reduce differentials in cancer prevention and cancer outcomes between people with intellectual disabilities and the general population.

Experts by experience and involvement

Collaboration is the key to success. People with intellectual disabilities across individual countries are often too small in number to have a strong impact on national policy. Recognising the importance of cooperation, knowledge, and experience exchange among experts in the field, the COST CUPID Action has established a network of academics, representatives and advocacy groups for people with intellectual disabilities to work together and propose measures to reduce health inequalities for these people in Europe and beyond. Also, the involvement of the intellectual disabilities community within different advocacy and non-governmental organizations as well as representatives of the World Health Organisation - working on cancer prevention and disability inclusion - should provide a stronger voice to be heard across Europe and globally, to promote equality of access to healthcare systems.

Inclusive research, as defined by Walmsley and Johnson ([Walmsley and Johnson, 2003](#)), is a research approach that involves people “who may otherwise be seen as subjects for the research as instigators of ideas, research designers, interviewers, data analysts, authors, disseminators and users” ([Nind, 2017](#)). Considering different methodologies and the involvement of people with intellectual disabilities, their roles in the inclusive research can span from advisory to active leadership roles with different degrees of support ([Strnadová et al., 2016](#); [Rojas-Pernia and Haya-Salmón, 2022](#)). This type of research improves the quality of data and increases the impact of the study findings to better address the specific healthcare needs of people with intellectual disabilities ([Frankena et al., 2019](#)). Inclusion of the people with intellectual disabilities is important for achieving change to the existing policy through advocacy. Investigating issues directly affecting people with intellectual disabilities and taking into considerations their views, knowledge and

experiences, can indicate directions for translating research findings into actions to improve the quality of their lives (Johnson, Minogue and Hopklins, 2014; Chalachanová et al., 2020).

Following the principles of inclusive research and ‘citizen science’, the CUPID Action will ensure the inclusion of people with intellectual disabilities, their families/advocates and caregivers as research partners in the policy evaluation process and co-production of knowledge for the development of pan-European cancer screening recommendations responsive to the unique needs of people with intellectual disabilities, which can further lead to the improvement in healthcare system (COST Association, 2022). It will include discussions via remote teleconferences, in person and/or written communication to understand factors which affect cancer prevention efforts and participation in cancer screening programmes for people with intellectual disabilities in order to provide evidence to serve relevant stakeholders and influence policy. This should produce a strong knowledge database benefiting from the experience of people with intellectual disabilities who will be significant stakeholders in the work process and reflecting a meaningful citizen science commitment of the CUPID Action, translating it into actions necessary to improve cancer-understanding prevention efforts in people with intellectual disabilities. This cooperation between interdisciplinary stakeholders and the community of people with intellectual disabilities will have an impact on a broader societal level, leading to cancer prevention and screening policy discussions, and health service reform.

Additionally, the CUPID Action brings together not only the expertise of its members but also their professional networks and contacts in the field, for the long-term collaboration and support in the future, especially for developing countries.

Conclusions

People with intellectual disabilities are often underrepresented in cancer prevention and screening policies or have limited access to these services, both of which lead to health disparities and unfavourable health outcomes in this population. The compelling evidence we discussed here underscores the critical need for interventions at policy and prevention levels, rather than solely at the individual level, for addressing the health disparities faced by people with intellectual disabilities in cancer prevention and screening. By influencing, or changing policy at a societal level, this may lead to wider changes, at a community, organisational and individual level, due to the interplay of these factors (Centers for Disease Control and Prevention (CDC), 2023). As such, the COST CUPID Action, and in particular the activities of the WG2, will generate a knowledge base of the best evidence, prevention strategies and screening practices, and will support the development of policy recommendations tailored to the unique needs of people with intellectual disabilities; this should have a positive long-term impact on the quality of life of this population. With the focus on EU policies of cancer prevention and the challenges of access to screening programmes, the CUPID WG2 will systematically collect data using a scientifically based approach, allowing direct comparative analysis of these policies for people with intellectual disabilities. Based on the acquired evidence and examples of good practice, it will provide recommendations for improvement to policy makers across Europe. The CUPID Action participants will benefit from interdisciplinary knowledge exchange and gaining new research skills during the lifetime of the project. By stimulating collaboration between like-minded people and organizations and using an interdisciplinary approach, it will build capacity to address the issues of health inequalities for people with intellectual disabilities. Ultimately, this will increase the network’s visibility and have a stronger impact on stakeholders and policy makers, which is particularly important for smaller and EU-

candidate countries, translating these efforts into a sustainable impact on cancer prevention and equity of access to screening.

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