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Priorities on rare cancers' policy in National Cancer Control Plans (NCCPs): A review conducted within the framework of EU-JARC Joint-Action

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ABSTRACT

Objectives: Although one out of four persons affected by cancer in Europe has a rare cancer (RC), it is unknown to which extent they are addressed in National Cancer Control Plans (NCCPs) and National Rare Diseases Plans (NRDPs). The objective of this review was to analyse the content included in European NCCPs and NRDPs on RCs in adults. The working hypothesis is that RCs have a lower presence in NCCPs compared to more common cancers, and that NRDPs do not generally approach them.

Methods: A review based on a documentary analysis on the priorities and recommendations in the area of RCs was conducted in 15 European NCCPs and 18 NRDPs. After identifying the areas covered therein, we performed a thematic analysis to allow a narrative description of the status of RCs' health policy.

Results: Rare cancers are hardly addressed in NCCPs and not addressed in NRDPs. Of the 15 NCCPs analysed, only 8 contained some elements on RCs, and only 3 of these described specific measures to address this disease group or took a comprehensive approach. The cross-cutting analysis of the 8 NCCPs allowed identifying 14 critical issues necessary to reach a comprehensive approach to RCs' policy.

Conclusions: The scarce presence of RCs in most of NCCPs may indicate low visibility and limited political understanding of their specificities. The critical issues emerging from the analysis are intended to improving the national policy frameworks addressing RC challenges and to place the NCCPs as strategic documents that must play a key role in this process.

1. Introduction

There are around 200 different types of rare cancers, including rare adult solid tumours (e.g. sarcomas, head and neck cancers, neuroendocrine tumours, central nervous system tumours), rare haematological cancers as well as all childhood cancers [1]. While they are a heterogeneous group of diseases, they share similar problems due to their rarity: uncertainty of diagnosis, lack of therapies, poor research opportunities, difficulties in clinical trials, and of centres of reference [2]. For children aged up to 14 years, cancer is the second most frequent cause of death and the first one by disease in children above one year. Together these cancers comprised the 24 % of the total cancer cases diagnosed every year in the EU-28 [3].

Against this background, the Joint Action on Rare Cancer (JARC) [4] was launched in 2017 to integrate and maximize efforts of the European Commission, MS and all stakeholders to advance quality of care and research on rare cancers. For the first time, JARC set forward the peculiarities of rare cancers versus rare diseases and cancer as such. In this line, it is also worth referring the establishment of the 24 European Reference Networks (ERNs), virtual networks involving healthcare providers across Europe [5]. Several ERNs are devoted to rare cancers: EURACAN for rare solid tumours in adults, PaedCan for paediatric cancers and EuroBloodNet for rare haematological diseases.

The WP10 of the JARC aimed at proposing a core set of strategies and measures to accommodate in the National Cancer Control Plans (NCCPs) and National Rare Disease Plans (NRDPs) across the EU Member States (MS). This study corresponds to the objective of addressing the specific needs of rare cancers in order to improve health outcomes through improved quality of care, better patient access to care, and reduction of inequalities across Europe [6].

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NCCPs and NRDPs are the expression of the priorities and measures proposed by policy makers to cope with the challenge of cancer and rare diseases, respectively [7]. This paper addresses the NCCPs and the NRDPs of 15 and 18 EU MS, respectively, to identify and analyse the content on health policy for rare cancers in adults included within the national cancer strategies and rare diseases plans, as well as to build upon the plans of MS a shared vision of the policy priorities and critical healthcare elements. The working hypothesis of this research is that rare cancers have a lower presence in NCCPs compared to more common oncological diseases, and that NRDPs do not generally approach them.

2. Materials and methods

2.1. Design

We performed a review based on a documentary analysis on the information, priorities, actions and recommendations in the area of rare cancers in different European countries. Primary documents were the National Cancer Control Plans (NCCPs) and the National Rare Disease Plans (NRDPs) of the European MS.

2.2. Document retrieval

As regards NCCPs, our document search benefited from previous work that identified cancer plans during the EPAAC (European Partnership for Action Against Cancer) and CanCon (Cancer Control) EU Joint Actions [8,9]. Two relevant sources of information for retrieving cancer plan documents were the EPAAC website [10] and the International Cancer Control Partnership (ICCP) portal [11]. The 2016 survey on NCCPs carried out in EU in 2016 as part of the CanCon initiative was also consulted [12]. The report describes the various terms used to refer to cancer plans, including 'programme', 'plan' and 'strategy'. Although this heterogeneity also extends to some degree to the contents of the documents, we will refer to them collectively (and synonymously) in this report as NCCPs or cancer plans. For its part, the NRDPs analysed were consulted at the Europlan Project' and EURORDIS' websites [13,14].

Following the data collection process, we examined only the NCCPs and NRDPs written in languages that our research team could comfortably work in or which were translated to English (see *Supplemental file*). Countries with included NCCPs and/or NRDPs are detailed in Table 1.

2.3. Analysis

NCCPs data were examined inductively, using content analysis to identify the 10 thematic areas that the NCCPs cover as well as the categories of analysis [15,16]. We summarised these areas in Table 2. This identification enabled the extraction and reorganisation of data regarding the content of the plans along each of those areas. Data were tabulated by country and section of the corresponding NCCP to facilitate subsequent access (see *Supplemental file*). Following the content identification, we performed a thematic analysis, grouping data into 5 topics to allow a narrative description of the status of rare cancers in European NCCPs (Table 3).The analysis also allowed identifying the extent to which plans contained information on rare cancer control (Table 4). These levels of information resulted in the following categories: 1) *no reference to rare cancers*; 2) *information covers one or two areas*; and 3) *information covers some areas*.

The reporting of results follows the priorities and recommendations made in each NCCPs; these are tagged with the official alphabetical country-code: Austria (AT), Belgium (BE), Czech Rep (CZ), Estonia (EE), France (FR), Germany (DE), Ireland (IE), Italy (IT), Luxembourg (LU), Malta (MT), Netherlands (NL), Portugal (PT), Slovenia (SI), Spain (ES), Sweden (SE), and UK-England (UK). The analysis concluded by identifying well-developed priorities and recommendations in the area of rare cancers.

Table 1
NCCPs and NRDPs included in the analysis in relation to all EU Member States and year of publication.

Country	NCCP (publication year)	NRDP (publication year)
Austria	X (2014)	X (2014-18)
Belgium	X (2008-10)	X (2015)
Bulgaria	–	X (2009-13)
Croatia	–	X (2015-20)
Czech Republic	X (2012)	X (2010-20)
Estonia	X (2007-15)	–
Finland	–	X (2014-17)
France	X (2018–2022)	X (2010-14)
Hungary	–	X (2013–2020)
Ireland	X (2017-26)	X (2014–2018)
Germany	X (2012)	X (2013)
Italy	X (2011-13)	X (2013-16)
Luxembourg		X (2018-22)
Malta	X (2017-21)	–
Portugal	X (2017)	X (2015)
Romania	–	X (2014–2020)
Slovak Republic	–	X (2016–2020)
Slovenia	X (2010-15)	X (2011)
Spain	X (2010)	X (2014)
Sweden	X (2009)	–
Netherlands	X (2005-10)	X (2017)
UK	X (2015-20)	X (2013)
UK-England	–	X (2018)
UK-N. Ireland	–	X (2015)
UK-Wales	–	X (2015)
UK-Scotland	–	X (2014)

3. Results

Of the 15 cancer plans we analysed, 8 considered rare cancers in adults to some extent, while 7 contained no information. Regarding the rare diseases plans, no information related to rare cancers was found. After identifying the cancer plans that contained information on rare cancers, we analysed the content to identify the cancer control areas covered (Table 2). In Table 3, we have reorganized these areas under five broader topics to facilitate the analysis and presentation of results. Table 4 summarises the extent to which this topic is developed in the cancer plans, according to three categories.

3.1. Epidemiological data and link to the rare diseases field

Five countries provided details on the incidence of rare cancers in their populations (IT,IE,MT,UK,SI), three of which (IT,IE,MT) referred to the lack of an internationally accepted definition for this group of diseases. NCCP authors highlighted the contrast between the definition of rare diseases based on prevalence set by EU Regulation (EC) No 141/2000 on orphan medicinal products (no more than 5 per 10 000 persons in the EU) and the threshold set by RARECAREnet (i.e., incidence of less than 6 per 100,000 population per year). The latter threshold would situate the incidence of rare cancers in different European countries from around 15%–22%. Furthermore, two cancer plans (IT,MT) subclassify rare cancers by the population group affected: rare adult solid tumours, rare haematological cancers and all childhood cancers (13 %, 8% and 1%, respectively for MT). There is also one plan that describes "very rare cancers" with an incidence established at < 1/100.000 pop./year (IT). These two plans (IT,MT) are the only ones that explicitly define rare cancers as rare diseases, highlighting the need to functionally integrate these two areas in the context of care networks (IT).

Table 2
Rare cancer control areas covered by NCCPs on rare cancers in adults.

COUNTRY	1.Definition of rare cancers and epidemiology	2. Linkage to rare diseases	3. Organisation of cancer services	4. Effective patterns of referral	5. Linkage to international centres of excellence	6. Histopathological and imaging diagnosis and early detection	7. Clinical research	8. Patients' involvement and availability of information	9. Evidence assessment and access to orphan drugs	10. Population-based databases, registries, biobanks
Austria	—	—	—	—	—	—	—	—	—	—
Belgium	—	—	x	—	—	—	—	—	—	—
Czech Rep	—	—	—	—	—	—	—	—	—	—
Estonia	—	—	—	—	—	—	—	—	—	—
France	—	x	x	x	x	x	x	x	—	—
Germany	—	—	—	—	—	—	—	—	—	—
Ireland	x	—	x	x	x	—	—	—	—	—
Italy	x	x	x	x	—	x	x	x	x	—
Malta	x	x	—	x	x	x	x	x	x	x
Netherlands	—	—	x	—	—	—	—	—	—	—
Portugal	—	—	—	—	—	—	—	—	—	—
Slovenia	x	—	x	—	—	x	—	—	—	—
Spain	—	—	—	—	—	—	—	—	—	—
Sweden	—	—	—	—	—	—	—	—	—	—
UK-England	x	—	x	—	—	—	x	x	x	—

Note: "X" signifies explicit mention in cancer plan; "—"indicates that the area was not covered.

3.2. Heath care organisation and quality

The quantity and variety of information on health services administration across different NCCPs is significant, as is the set of strategies intended to improve access to high-quality care in a context requiring increasing multi-level coordination (hospitals with different levels of complexity, primary care, etc.). For instance, rare cancer care is denominated 'quaternary care' (IE) due to the high level of specialisation required and the need to reorganise services to improve the diagnostic and therapeutic approach. Changes are described at all levels: responsibilities for this type of patients within expert teams, changes in decision-making processes at the geographic level (among others), and infrastructure requirements. From this macro perspective, another plan (UK) alludes to the necessity of commissioning the services dedicated to these pathologies at the national level, together with paediatrics, adolescent health and young adult health. Both perspectives share the idea of establishing a specific policy framework for rare cancers that enables improvements in their control and the related care.

A common measure found in NCCPs is centralising case management or (from a similar perspective) to identify centres of excellence (IT,IE,SI,NL,BE,FR). The explicit logic for this strategy resides in the need to offer patients with rare cancers the best clinical expertise available, and indirectly to improve professional specialisation within the health system by increasing the volume of cases handled in specific centres. The plans also mention the need to seek economies of scale with regard to high-tech resources (IT) and the importance of setting qualitative as well as quantitative thresholds. The corollary is that the centres responsible for taking care for patients with rare cancers should also be responsible for the resources (including the specialised human resources) and organisation needed to provide it (BE). The concentration of rare cancer cases in expert centres is the preeminent strategy for these diseases in cancer plans.

Another element that also stands out in several NCCPs is the role that expert multidisciplinary teams (MDTs) should play at the national level (IE,UK,SI,NL,BE,FR). Two cancer plans highlight the need for cancer networks to facilitate access to such teams (FR,IT). In that sense, a distinction can be made between the countries where certain teams will take on all cases (IE), for example for soft tissue sarcoma or neuroendocrine cancer, and the countries with two broad levels of expertise: one at the regional and one at the national level. According to the latter model, some teams may be accredited to carry out diagnostic and treatment services for some rare cancers, but they must validate the treatment strategy with highly specialised teams of experts or directly refer patients to these teams if the complexity of the case crosses an established threshold (FR,UK). This difference may be related to the population size.

Thus, the policy of centralisation lies at the intersection between the need to have expert teams and the decision on how much to centralise services (then increasing volume and promoting specialization), with the result that there may be 'expert teams', 'nominated physicians' (IE) in 'designated centres' (IE), 'centres of excellence' (IT), 'tertiary institutions' (SI) or 'centres of expertise' that assume these cases.

Two additional main elements that NCCPs include as key measures when organising rare cancer services are continuity of care and effective patient referral. Continuity of care entails, for example, the need to specify the professionals and tasks in the coordinated care chain,

Table 3
Rare cancer control areas covered by NCCPs, according to five broad topics.

Cancer control areas covered by NCCPs	Topics
1. Definition of rare cancers and epidemiology	**(a) Epidemiological data and link to the rare disease field**
2. Linkage to rare diseases	
3. Organisation of cancer services	**(b) Healthcare organisation and quality**
4. Effective patterns of referral	
5. Linkage to international centres of excellence	
6. Histopathological and imaging diagnosis and early detection	**(c) Clinical practice and research**
7. Clinical research	
8. Patients' involvement and availability of information	**(d) Patients' involvement and availability of information**
9. Evidence assessment and access to orphan drugs	**(e) Health Technology Assessment (HTA) and data registration**
10. Population-based databases, registries, biobanks	

Table 4
Levels of information on rare cancers in NCCPs.

Country	Categories	*a) Epidemiological data and link to the rare disease field*	*b) Healthcare organisation and quality*	*c) Clinical practice and research*	*d) Patients' involvement and availability of information*	*e) HTA and data registration*
Austria	1	–	–	–	–	–
Czech Republic		–	–	–	–	–
Estonia		–	–	–	–	–
Germany		–	–	–	–	–
Portugal		–	–	–	–	
Spain		–	–	–	–	–
Sweden		–	–	–	–	–
Belgium	2	–	X	–	–	–
Netherlands			X	–	–	–
Slovenia		–	X	–	–	–
Ireland		X	X	–	–	–
UK-England	3	X	X	X	X	X
France		X	X	X	X	–
Italy		X	X	X	X	X
Malta		X	X	X	X	X

Note: 1 corresponds to "no reference to rare cancers", 2 to "information covers one or two areas", and 3 to "information covers most of areas".

including the GP, extramural carers and hospitals (when necessary) (NL). The identification of tasks in hospitals is an essential condition for the effectiveness of procedures, logistics and communication functions between hospitals or with the patients themselves. In that line, another cancer plan recognises geography as a challenge for coordinating care in patients with rare cancers, proposing the development of 'cancer specialist nursing roles' as one measure to address it (UK).

As for effective patient referral, planners describe in one NCCP the need to establish clear pathways for the diagnosis and treatment of rare cancers, which implies easy access – and timely transfer of care – to reference centres and MDTs (IE). Another plan states that the identification of centres of excellence should contemplate referral in the context of patient migration (IT). Patients' freedom of choice with regard to centre and across different healthcare areas should be underwritten by the reimbursement mechanisms that permit it (MT,IT); likewise, barriers to choice (such as waiting lists) should also be minimised (IT). Another cancer plan highlights the importance of rapidly managing patients, guaranteeing that they are treated with the level of complexity they require and of approaching any individual condition with an adequate response at regional or interregional level (FR). The objective, as stated, is to ensure that patients do not miss any opportunities for the most adequate treatment (including innovative therapies) or services. Thus, the role that expert MDTs play in these decision-making processes is critical. Local clinical teams may be able to manage these types of patients, but the expert MDTs will be responsible for validating the proposed treatment strategy or for assuming care of the patient directly (FR).

Finally, three island countries (or countries with some island territory) highlight the importance of linkage to international centres of excellence for improving management of people diagnosed with different forms of rare cancers (IE,MT,FR). Planners argue that the transfer of specialist knowledge and expertise should include cross-border centres, including through participation in ongoing activities at EU level in the field of rare cancers (MT). This change should include the establishment and maintenance of contacts and communications with relevant experts based on instruments that facilitate connectivity, for instance telemedicine, digital pathology systems or international centres of excellence. Some of these statements have been made previously or in parallel to the creation of the European Reference Networks (ERNs).

3.3. Clinical practice & research

Diagnosis and clinical research for rare cancers are two key elements within cancer plans. Early detection and diagnostic processes are critical in the field of rare cancers, and four cancer plans emphasise this point in order to improve patient access to the maximum range of treatment options (FR,IT,SI,MT). In that sense, one measure that stands out is the facilitation of double readings at pathological and image level. Errors in histopathological diagnoses are frequent in rare tumours, which should lead to a diagnostic review in centres of excellence or direct referrals to these centres for diagnosis (FR). Expert pathologists and radiologists should be based in these centres or have a priority role there in order to provide a high-quality service (FR). International collaboration should also be enabled through this approach (IE,MT).

Supporting double reading processes has led to the recommendation of certain measures to facilitate its implementation. Some plans have proposed specific mechanisms for reimbursement (IT,FR), while another plan recommends situating these processes within a general framework that is coherent with the care that patients with a rare cancer receive. In turn, this should occur while harmonising the organisation and financing of the devices that these patients assume (FR). In general, plans also emphasise the importance of centralising more complex diagnostic tests to favour the efficient distribution of resources (SI).

In the area of treatment and research, several cancer plans promote research into rare cancers, considering these fields to be "underserved" (IT,MT,UK,FR). The research can be performed in an academic context, using public funds to make up for the lack of research in the current pharmaceutical market (IT), or financing from industry partnerships (FR). Planners also mention the opportunity offered by the new EU Clinical Trials Regulation to reduce the time it takes to set up studies, which opens the door to additional clinical trials in the area of rare cancers. There are also generic references to the fact that the quality of care should be equivalent for every provider and that centralising diagnosis and the planning of treatment strategies in expert centres should be organised in line with the best international practice.

3.4. Patients' involvement

Generally, rare cancer patients report less satisfactory experiences in relation to care provided than patients with common cancers (UK). One critical aspect covered in different cancer plans has to do with the available information, as this can demand a greater effort on the part of the patients to find reference centres and specialists for diagnostic, treatment and post-treatment services. Considering that not all reference centres include the whole range of diagnostic and therapeutic procedures, their role in providing patients with comprehensive information is critical (IT,UK); reference centres should offer patients a directory of services, with signposts for how and where to find the most appropriate specialists. Another issue highlighted is the importance of involving the patients' communities (IT) and using patient-reported outcome measures (PROMs) and evaluations of care experiences to

enrich findings from clinical research, thus amplifying patients' perspectives and priorities (MT).

3.5. HTA and data registration

The specificity of rare cancers has led some NCCPs to introduce relevant considerations on the assessment of available evidence, particularly given the implications that this might have in terms of patient access to drugs or other therapies (IT,UK,MT). Avoiding discrimination against this patient profile may entail not applying the same quality standards to evidence evaluation in the decision-making processes around indications, which could result in a higher degree of tolerance of risk-adverse approaches. In this line, there are proposals for methodological innovation for adapting the biostatistical concepts of validity and precision to the circumstances of rare cancers (IT). As a corollary, the conditions for using drugs in Phase II studies ('compassionate use') should be relaxed even if there is only partial evidence of positive outcomes and an international consensus exists. A further issue covered is the need to protect access routes to drugs for rare cancer patients (MT). Finally, one cancer plan sets the objective of collecting specific population-based information on diagnosis and treatment of rare cancers (MT), lamenting the scarcity of registries and tissue banks for these pathologies.

4. Discussion

Our results indicate that despite their contribution to the overall cancer incidence, rare cancers are not a prominent topic in NCCPs, the principal instrument used by European countries to organise their cancer services. Of the 15 cancer plans analysed, only 8 contained some information on rare cancers, and only 3 of these included specific measures to address this disease group across the different areas of cancer control ("Healthcare organisation", "Patients' involvement", etc.). While the content on priorities, recommendations and best practices is significantly variable between countries, the cross-cutting work resulting from the analysis allowed overcoming the country-based perspective and integrating the relevant contents in the following priorities and critical points:

4.1. Quality of care and health care organisation

1 Centralising care for patients with rare cancers in reference centres emerges as a necessary condition for effecting change in the organisation of services at different levels, especially: personalising care, having updated clinical protocols, improving professionals' clinical competencies, assessing care quality in health centres, increasing patients' participation in clinical trials, and improving the conditions for research and development on new therapies.

2 Care for patients with rare cancers should be based on expert MDTs, which should in turn be articulated with other levels of care. The patient's reference centre needs to be fully coordinated with other expert centres at national and/or international level, avoiding silo models. Centralisation should not impede the fluidity of knowledge exchange between professionals and specialised centres.

3 The possibility of treating a patient with rare cancer (e.g. sarcoma) in one centre should not prevent collaboration with other centres with greater expertise in the case of a pathological subtype (e.g. bone sarcoma) or particular clinical condition. In these cases, administrators should facilitate the transfer of knowledge so that the anatomopathological diagnosis and/or treatment plan is validated with the highest available level of expertise, or that the case is directly transferred to the most expert centre.

4 Continuity of care is a critical dimension. The health system should manage the possible changes in centres, services and reference professionals derived from patients' changing needs and difficulties in access due to geographic distance. Team leaders or other professionals with a specifically designated role should manage transition points, for example referrals to expert centres or a patient's decision to change centre. A networking approach based on a 'hub and spoke' model can contribute to rationalise the health care migration, particularly when treatments have been centralised in reference centres.

5 Centralising patients in expert centres, combined with quick referrals for services therein and a financing system that does not disincentivise the practice, is a key way to promote equity, in that patients will not lose the opportunity to access the maximum range of treatment options, including innovative therapies.

6 Avoiding errors in anatomopathological diagnosis is crucial. In a context of centralised care, special consideration should be made of guaranteeing high-quality anatomopathological diagnosis and being equipped with high-tech laboratories for performing molecular diagnosis. Reducing mistakes in first diagnosis through centralisation and medical education is critical. However, systems for double reading should also contribute to this measure, making it relevant to adapt the organisation of services and centres in line with the objective of increasing clinical safety and guaranteeing maximum equity in the diagnosis. This should be encouraged through adequate funding.

7 It is important to help patients obtain a second medical opinion when desired, without necessarily breaking ties with the reference care team. Normalising this situation is relevant for clinical safety and for reducing the distress that patients and families may feel.

8 Telemedicine and the use of digital pathology systems can be normalised in order to improve the connectivity between centres that treat patients with rare cancers, thus ensuring the transfer of expert knowledge.

4.2. Research

9 Clinical research and the development of new treatments are considered 'underserved' in this area. Accelerating development of new clinical trials is a priority, but so is incorporating other methodological and research perspectives, for example, tissue analysis for understanding the molecular characteristics of cancer in the development of new therapies; relaxing some conditions in the evaluation of evidence in order to indicate treatments; perform academic clinical trials; and launch public-private partnerships. In this context, accelerating and strengthening biobank networks is essential to enable the validation of prognostic factors and develop new treatments.

4.3. Patient involvement

10 The drive to improve care and research into rare cancers requires amplifying the patient's perspective. Involving patients when establishing priorities for clinical research and service provision can be articulated, for example through processes to collect patients' and their families' experience and feedback for the use of PROMs.

11 Based on the model of NRDPs in Europe, where rare disease patient representatives are involved in the design, development and implementation of a RD Plans, the patients with rare cancers and their representatives must be involved in the design and drafting process of NCCPs on matters related to rare cancers, due to their intimate knowledge of the diseases, their experience in research projects and healthcare policies to enable a better access to required multidisciplinary specialised healthcare services.

4.4. Stakeholder involvement

12 Health authorities cannot simply be 'one more' actor in the area of rare cancers. Rather, their role should be very active, especially in establishing quality criteria for services, designating and

consolidating reference centres, coordinating providers and improving research conditions.

13 The third social sector (non-profits) can play an important part in meeting some of patients' necessities.

14 The centres that treat patients with rare cancers should be the main source of information for patients on the most adequate range of specialists and services for their care.

4.5. International collaboration/EU perspective

15 Due to the rarity of each single disease, rare diseases and rare cancers have a strong European added value as no one country alone can tackle the issue of both RDs and RCs. Since European regulations, policies and recommendations have been developed to address the challenges paused by the rarity of a disease, it is of importance that EU MS integrate relevant European policies for rare cancers in their NCCPs. In this line, the national expert centres who are members of ERNs covering rare cancers, and the connection between these ERN members and other healthcare professionals/national networks should be supported at the national level to optimise the offer for care.

This study has some strengths and limitations. The main limitation is that we were not able to include all EU cancer plans in the analysis. First of all, not all countries have published their plan, which limited the availability of the documents to us. Moreover, each plan is written in the national language(s) of the country, but its translation to English is infrequent. At the same time, we excluded plans published before 2008, as we assumed they were no longer in force. In some cases there have been substantial changes even recently. For example, in Italy the rare tumours network was formally separated in 2017 from the network of rare diseases, with the former only depending on cancer centres. It is also worth noting that some regional plans exist, but our analysis was limited to those at a country level. In addition, we included information exclusively related to rare cancers, that is, we did not formally consider how services for these cancers may have been influenced by policies or cross-sectional measures developed for all oncological diseases. Also, contents related to paediatric cancer were excluded as we aimed at focusing on rare cancer in adults. Strengths of the study were the validation of results, carried out by researchers from different institutions, and the systematic process of data analysis, as it can be directly traced from the original sources in the supplemental file.

Aside from methodological limitations, it should be noticed that even if the cancer plans analysed are in force or were recently approved (Table 1), most of them do not reflect the initiatives that have taken place in recent years at European level. For instance, with the creation of ERNs, for the first time, an EU response has been formulated in an area that has traditionally been under the exclusive power of MS. The reason is that the principle of subsidiarity [17], which rules out Union intervention when an issue can be dealt with effectively by MS at central, regional or local levels, is dysfunctional in the field of rare diseases. The low volume of patients and the fact that clinical expertise on the different pathologies is unevenly distributed among the states implies that only a shared framework may benefit all EU citizens. Also, the EU Joint Action JARC was launched in 2017 as a framework to prioritise rare cancer in the agenda of the EU and cancer plans at National level and develop innovative and shared solutions, mainly to be implemented through the future ERNs on rare cancers in areas such as quality of care or diagnosis and treatment. JARC and ERNs combined represent a concrete opportunity to make networking a reality, reduce disparities and improve outcome in these diseases [18].

In conclusion, while rare cancers comprise almost 1 out of 4 cancer cases diagnosed every year in Europe, they are hardly addressed in national cancer plans. It is crucial to strengthen their presence therein as a first step to increase their visibility and political commitment. This research resulted from a collaborative effort between research institutions, governmental bodies and patient associations, and intended to contribute to this process by taking together all of the EU cancer plans and fostering a more comprehensive, European approach to rare cancer care. While a number of lessons can be drawn the world of rare diseases, the rarity of rare cancers should be embedded in NCCPs as umbrella instruments of the set of policies and measures to be undertaken in cancer care. The policy and organisational priorities emerging from the analysis might help improving the national policy frameworks for these cancers in a complementary and synergistic way to the ERNs' expansion.

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Declaration of Competing Interest

None.

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Appendix A. Supplementary data

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