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ORIGINAL RESEARCH ARTICLE

Developing a Conceptual Framework for Socioeconomic Impact Research in European Cancer Patients: A 'Best-Fit' Framework Synthesis

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Abstract

Background Multiple studies have indicated a socioeconomic impact of cancer and cancer care on patients and their families. Existing instruments designed to measure this impact lack consensus in their conceptualization of the issue. Further, various terminologies have been used in the literature (e.g., financial burden, financial hardship, financial stress) without clear definitions and consistent conceptual background. Based on a targeted review of existing models addressing the socioeconomic impact of cancer, our goal was to develop a comprehensive framework from a European perspective.

Method A 'best-fit' framework synthesis was applied. First, we systematically identified existing models to generate a priori concepts. Second, we systematically identified relevant European qualitative studies and coded their results against these a priori concepts. Inclusion and exclusion criteria were predefined and applied thoroughly in these processes. Thematic analysis and team discussions were applied to finalize the (sub)themes in our proposed conceptual framework. Third, we examined model structures and quotes from qualitative studies to explore relationships among (sub)themes. This process was repeated until no further change in (sub)themes and their relationships emerged.

Result Eighteen studies containing conceptual models and seven qualitative studies were identified. Eight concepts and 20 sub-concepts were derived from the included models. After coding the included qualitative studies against the a priori concepts and following discussions among team members, seven themes and 15 sub-themes were included in our proposed conceptual framework. Based on the identified relationships, we categorized themes into four groups: causes, intermediate consequences, outcomes and risk factors.

Conclusion We propose a Socioeconomic Impact Framework based on a targeted review and synthesis of existing models in the field and adapted to the European perspective. Our work contributes as an input to a European consensus project on socioeconomic impact research by an Organization European Cancer Institute (OECI) Task Force.

Key Points for Decision Makers

There is a large variety in how the socioeconomic impact of cancer is conceptualized and measured.

In this study, we integrate conceptual models and observations of how people are socioeconomically affected by cancer.

The study is part of a larger OECI Task Force in which a comprehensive conceptual framework is developed for the assessment of the socioeconomic impact of cancer.

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1 Introduction/Background

Cancer patients and their family members may face out-of-pocket (OOP) costs or income/work loss due to cancer treatments and medications, related symptoms and subsequent adverse effects [1–3]. These impacts of cancer and cancer care have become a topic of concern in the US [3–5], and the concept of 'financial toxicity' has been widely used to "describe how out-of-pocket costs can cause financial problems for a patient" [5]. OOP costs refer to the costs that patients have to pay for their medical care that is not covered by their health insurance [5].

These impacts of cancer and cancer care are not limited to the costs of medical care, but can also stem from income/work loss as a result from cancer and its treatments [6]. Patients' family members may be affected by these impacts as well [7, 8]. A variety of terminologies have been used in existing studies to describe this phenomenon, for example, financial burden, financial hardship, financial stress, economic burden, among others, without a clear definition and conceptual background [2, 3, 9].

Regarding measurement, instruments have been used to address the financial toxicity of cancer from a patient's perspective. One example is the COmprehensive Score for financial Toxicity (COST) [10, 11]. This instrument was originally validated using a sample of advanced (stage IV) cancer patients (unspecified cancer types) in the US and was intended to measure financial toxicity as a single concept. More recently, the Financial Index of Toxicity (FIT) instrument, validated using a sample of head and neck cancer patients in Canada, identified three sub-concepts under the concept of financial toxicity: financial stress, financial strain and lost productivity [12]. As with the definitions, these two instruments, among others, vary in the number and type of concepts or sub-concepts utilized, with the intention of measuring the same phenomenon, that is, 'financial toxicity'. These differences in the conceptualization of financial toxicity could be attributed to variations in healthcare systems between the US and Canada. Canada has a publicly funded national health insurance system, while the US primarily relies on private financing and delivery [13]. Although Canada's health system can be considered to be similar to those in Europe regarding commitment towards universal health coverage [13, 14], there is significant heterogeneity among European countries, for example, in terms of health financing [14] or accessibility to healthcare [15]. Despite the heterogeneity, in this study, we generally consider the European context to encompass those welfare states with a comprehensive universal healthcare system in Europe.

To address this diversity in conceptual background, terminology and context, we aim to review available conceptual models in the research area of social and economic (socioeconomic) impact (SEI) of cancer and to adapt relevant concepts to the European perspective. Accordingly, we conducted a best-fit framework synthesis, a process that combines both framework and thematic analysis techniques, to (i) review available concepts in existing conceptual models/frameworks and (ii) synthesize the qualitative studies from a European context addressing the SEI of cancer with the goal to develop a comprehensive conceptual framework of SEI that is particularly relevant to use by healthcare professionals, researchers or policy stakeholders in the European context. Our work contributes to a European consensus project on SEI research by an Organization European Cancer Institute (OECI) Task Force [16].

2 Methods

2.1 'Best-Fit' Framework Synthesis

We implemented a 'best-fit framework synthesis' approach [17, 18]. This technique is composed of framework synthesis and thematic analysis techniques to develop a best-fit conceptual framework in a particular research field and its context.

The process started with a targeted identification of relevant models as well as European qualitative studies (i.e. studies which collected data via in-depth interviews, focus group discussions, observations, etc. [19]) addressing the SEI of cancer (see Fig. 1). Next, all concepts from existing theoretical models were extracted and analyzed to generate a priori concepts and sub-concepts based on their commonalities and differences and their definitions.

In the following step, data from the results sections of included qualitative studies were extracted for thematic analysis against the a priori concepts and sub-concepts. This data consisted of the primary analysis of the authors as well as participant quotes appearing in the results sections of these studies. If any of the information did not fit to the a priori concepts and sub-concepts, henceforth referred to as (sub)concept(s), a new (sub)concept was generated using thematic analysis techniques. A consistent combination of a priori and new (sub)concepts was then discussed among the authors (PDP and JU) to finalize the themes and sub-themes, henceforth referred to as (sub)themes, to be included in our conceptual framework. Finally, we explored the relationships among (sub)themes to formulate our conceptual framework (see Fig. 1).

For more clarity of our synthesis process, specific terms (e.g., 'concepts' vs 'themes') used to describe how we developed our framework are presented in Table 1.

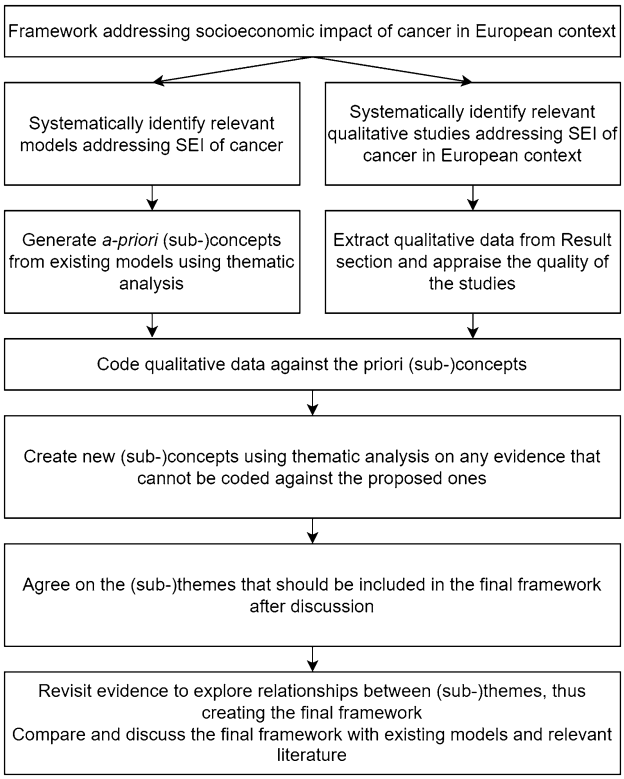

Fig. 1 'Best-fit' framework synthesis process (modified from [17]). *SEI* social and economic impact

2.2 Inclusion Criteria

The current study uses a sub-sample of articles from a database created for a systematic literature review that examined the current terminology used to refer to the SEI of cancer [9]. Briefly described, this systematic literature review used three search engines: PubMed, EconLit and Web of Science (WoS). The search was limited to articles published from January 1, 1979 to September 30, 2020, and only articles in English were included. Search strategy and selection process are summarized in Supplementary Material 1 of the electronic supplementary material (ESM). This database included 595 articles in total. The current study utilized this database and selected articles with the following inclusion criteria: (i) studies with existing models related to SEI of cancer (i.e. models of financial toxicity, financial impact, financial burden, etc.); and (ii) qualitative studies that related to the SEI of cancer in the European context for the best-fit synthesis.

2.2.1 Identification of Existing Models

All records from the above-referenced database were further processed by conducting two parallel search strategies to ensure that all existing models were captured.

1. The search strategy was based on recommendations by Carroll et al. [17]. We excluded articles if the title/abstract did not contain at least one of the following terms: *model, theory, theories, framework, concept* or *conceptual*. Furthermore, we retained all systematic reviews in the database since they potentially contained desired models in the summary of their findings. Those remaining eligible studies were scanned and cross-reference checked to identify the studies containing models.
2. A quick scanning strategy. All the records in the database were scanned to identify models or frameworks related to SEI of cancer in their figures.

2.2.2 Identification of Qualitative Studies and Their Quality Appraisal

In order to identify the qualitative studies addressing SEI of cancer in the European context, the inclusion criteria were developed based on the published search strategy SPIDER, the key elements being **S**etting/population, **P**henomenon of **I**nterest, **D**esign, **E**valuation, **R**esearch [20]. Accordingly, the following inclusion criteria were used:

- *Setting/population*: European countries
- *Phenomenon of Interest*: Socioeconomic impact of cancer

Table 1 Clarification of terms used in this study

Term	Clarification
Synthesis	The general term for the whole 'best-fit framework synthesis' process in this study
Thematic analysis	Qualitative analysis method used to analyze a priori (sub)concepts and data from included qualitative studies
Model	The models/frameworks/theoretical concepts from previous studies that are related to the socioeconomic impact of cancer (18 models in this study)
Conceptual framework	Our proposed model that is the result of the synthesis conducted in this paper (the Socioeconomic Impact Framework)
(Sub)concept	The concept and/or sub-concept derived from existing evidence, either included models or qualitative studies
(Sub)theme	Theme and/or sub-theme included in our conceptual framework, after thematic analysis

- *Design, Evaluation, Research*: Data collected using qualitative methods (e.g. in-depth interviews, focus groups discussions, observations) [19]

Identified studies were appraised using the consolidated criteria for reporting qualitative research (COREQ) [21] with a 32-item checklist by two research members (PDP and JU).

2.3 Data Extraction

2.3.1 Existing Models

Existing models that satisfied the inclusion criteria were included for the development of a priori concepts and sub-concepts. The names of the models, overall terminologies, concepts, sub-concepts and their definitions (when provided) were extracted. Afterwards, we conducted a thematic synthesis with these concepts and terminologies to create a list of a priori concepts and sub-concepts by consolidating the commonalities and generalizing the differences between the concepts in the included models. To avoid the possibility of overlooking relatively minor differences between identified models, we incorporated all of them in our analysis. The thematic synthesis was performed in Microsoft Excel qualitatively by the first author (PDP), available upon request.

2.3.2 Qualitative Studies

Two reviewers (PDP and JU) independently coded the qualitative data line by line against the a priori concepts and sub-concepts. For any data point that could not be accommodated in these a priori (sub)concepts, a new (sub)concept was generated. After coding was completed, the two reviewers discussed the results and identified and resolved inconsistencies in their respective categorizations. The process of coding was repeated until no additional (sub)concept was identified (3 rounds total) and until there was agreement regarding the additional (sub)concepts that emerged from the thematic analysis (performed by PDP and JU). These activities were performed in OpenCode 4.03 software for qualitative analysis [22].

2.4 Synthesis and the Formation of an Adapted Conceptual Framework

After discussion, the two reviewers agreed upon the final list of (sub)concepts for synthesis. The synthesis consisted of two stages. First, all a priori (sub)concepts from the existing models and those newly identified from the thematic analysis of the qualitative studies were listed. These (sub) concepts were defined clearly and included as themes and sub-themes for our proposed conceptual framework. Second, the evidence underpinning the (sub)themes was examined to discover the relationships among them and to develop connections in our framework. This information came from how the existing models were constructed and from statements in the included qualitative studies.

2.5 Sensitivity Analysis

We performed a sensitivity analysis for existing models in which five US-based models [3, 5, 23–25] and one model based in low- to middle-income countries [26] were excluded from the synthesis, as these healthcare contexts are different from Europe in regard to universal health coverage. We assessed whether these exclusions influenced the derivation of the a priori concepts from the remaining models. First, we evaluated whether any of the a priori (sub)concepts could have been omitted because of the exclusion of these models. Furthermore, we assessed whether the exclusion influenced the thickness of definitions of each of the (sub) concepts provided by the remaining models.

3 Results

3.1 Included Models and Qualitative Studies

We identified 12 models using the search strategy based on recommendations by Carroll et al. [17] and 14 models using the quick scanning strategy. After removing the duplicates, 18 models from 18 articles were included in this study. The PRISMA flowchart is presented in Fig. 2.

Seven qualitative studies addressing SEI of cancer in the European context were identified. Given this small number, we included all these seven studies for data extraction. The results of quality appraisal were discussed for consistency between two investigators (PDP and JU) and are summarized in Supplementary Material 2.1 (see ESM).

Descriptive characteristics of the included 18 models and seven qualitative studies are summarized in Supplementary Material 2.2 (see ESM); characteristics include first author, year of publication, type of publication, country(s), study population, type of cancer, age group and model structure.

3.2 A Priori Concepts Derived from Existing Models

Eighteen models in eighteen publications were included in the thematic analysis. The synthesis of these models is represented in Table 2. In this synthesis, terminologies were selected from previous models to define these a priori (sub) concepts. This synthesis resulted in 8 concepts and 20 sub-concepts. Table 3 provides a listing of these (sub)concepts along with an indication of whether the (sub)concept was included per each model. The definitions of a priori concepts

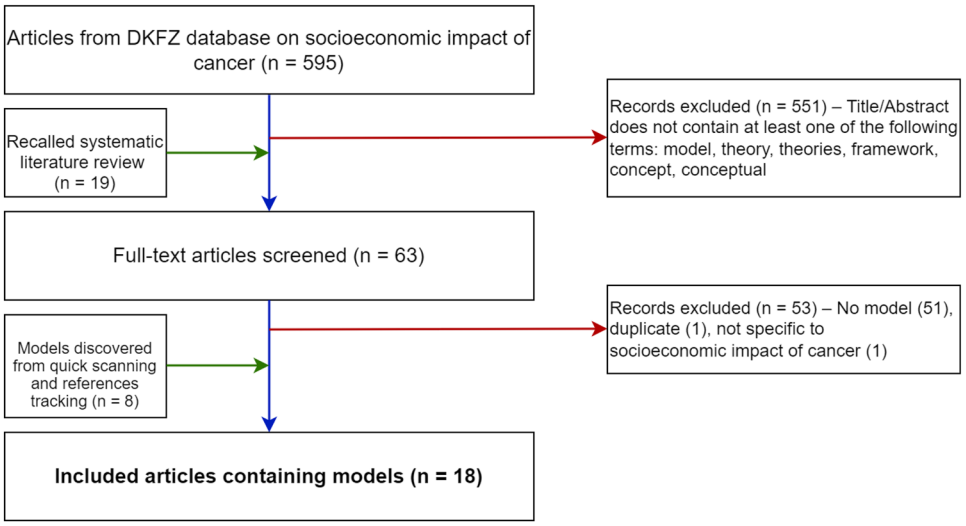

Fig. 2 PRISMA flow diagram—the systematic identification of existing models. *DKFZ* German Cancer Research Center

and sub-concepts are summarized in Table 4. Studies from which these definitions were derived are cited in the last column of this table. Each (sub)concept definition was derived from at least two publications.

The most common concept identified in the previous models was 'psychological response' ($n = 16$) and the least common was 'financial outcome' ($n = 5$). The average number of concepts per model is 4.28 (Table 3). Five concepts—'direct costs', 'indirect costs', 'financial coping behavior', 'psychological response' and 'risk factors'—contain sub-concepts.

Three important observations were made from the compilation of a priori (sub)concepts. First, regarding the definition of concepts in the identified models, the concepts in some of the models were unspecific [5, 27, 28]. These articles—all systematic reviews—summarized empirical studies and mainly identified which questionnaires measured those concepts, without defining them. Other models [2, 3, 24, 25, 29, 30] defined a concept by listing the included sub-concepts, then only defining the sub-concepts.

Second, we noted that there were two different definitions of a similar concept, 'objective measures'. Some authors [2, 31] defined 'objective measures' of financial burden of cancer as the tangible coping mechanisms, for example, using savings, selling assets, or borrowing money. Other authors [32, 33] regarded 'objective measures' as the direct and indirect costs of cancer care. Differences in definitions were also identified for the concept 'subjective measure'. In some studies [2, 32], authors only referred to the psychological impact of financial burden of cancer, while in other studies [31, 33], authors also included some of the coping behaviors in their definition. These differences may arise from the different perspectives on what 'objective' and 'subjective' is. We decided not to use these terms to prevent confusion and instead opted to develop different concepts that reflect each of these understandings (see Table 4).

Finally, most of the sub-concept definitions of the 'risk factors' were derived from Yabroff et al. [34], who specifically examined the associated factors affecting SEI of cancer on patients and their families.

3.3 The Formation of Our Conceptual Framework

After coding the information from qualitative studies in the European context against the a priori (sub)concepts and after resolving disagreement, we generated the themes, subthemes and their definitions for inclusion in our proposed conceptual framework (see Table 5). Supplementary Material 2.3 summaries the names and definitions of the a priori (sub)concepts derived from existing models and the (sub) themes of our proposed conceptual framework for comparison (see ESM). Further, we categorized the themes into four meta-themes: causes, intermediate consequences, outcome and risk factors. The hypothesized relationships between these meta-themes were based on the structures of included models and evidence from qualitative studies. This resulted in our proposed conceptual framework, shown in Fig. 3.

3.3.1 The Causes—Direct and Indirect Costs

3.3.1.1 Adjustment from the a priori concepts The names of two themes, 'direct costs (OOP expenditure)' and 'indirect costs (productivity loss)' and their respective subthemes, 'direct medical costs' and 'direct non-medical costs' (direct) and 'time loss at work' and 'income loss' (indirect), remained unchanged from the a priori concepts and were mentioned in all seven qualitative studies [35–41]. Authors used some form of the phrasing 'direct costs', 'direct non-medical costs' and 'direct medical costs', or their implications, substantially.

Table 2 A priori concepts derived from existing models

References	Direct costs	Indirect costs	Material resources	Psychological response	Financial coping behavior	Financial outcome	Health outcome	Risk factors
Hanratty et al. (2007) [28]	Rising health care costs and increasing health care use			Financial strain: the subjective perception of financial hardship				
Kankeu et al. (2013) [26]	Economic consequences: Direct costs	Economic consequences: Indirect costs	Social resources (e.g assistance from others)		Coping strategies (e.g. Intra- and inter- household labor substitution; Hiring other labor and other strategies; Reducing/delaying consumption of non-health goods and services (food, education, electricity, leisure, etc.); Use of savings; Sale of assets; Borrowing; Delaying investments)			Treatment seeking behavior: Yes or No
Gordon et al. (2017) [2]	Currency (monetary) values of OOP costs		Percentage of OOP costs to income ratios	Perceptions of cancer-related financial burden and psychological impact	Tangible solutions to ease financial burden such as increasing debt levels, borrowing money from family or friends, selling assets			
Altice et al. (2017) [3]	Material conditions: Increased OOP expenses	Material conditions: Lower income that can result from the inability to work during/following cancer treatment		Psychological response: Response to the increase in household expenses that must now be managed as patients navigate cancer care	Coping behaviors: patients adopt to manage their medical care while experiencing increased household expenses during/following cancer care	Medical debt/bankruptcy		
Santacroce et al. (2018) [29]	Monetary: Direct costs	Monetary: Indirect costs		Patient-reported: Psychosocial	Patient-reported: Financial coping			

Table 2 (continued)

References	Direct costs	Indirect costs	Material resources	Psychological response	Financial coping behavior	Financial outcome	Health outcome	Risk factors
Witte et al (2019) [33]	Objective financial burden: Direct costs (OOP)	Objective financial burden: Indirect costs (income loss)	Subjective financial distress: Material	Subjective financial distress: Psychosocial	Subjective financial distress: Behavioral			
Yabroff et al. (2018) [34]								Level of risk factors: patient/family, provider and care team, health care system, employer, state and national policy
Thomas et al. (2019) [27]	Direct (medical) costs, indirect (non-medical) costs			Biological responses to stress	Financial coping behavior: Treatment non-adherence			Medical insurance
Imber et al. (2020) [42]	Direct costs: Patient's OOP responsibilities owing to prostate cancer	Indirect costs: Opportunity costs of prostate cancer	Individual economic circumstances: Baseline wealth, pre-existing debts and economic reserve	Expectations of possible financial burdens: Knowledge and perception of the economic effect that a prostate cancer diagnosis will have on one's family				Patient-specific values and self-management behaviors
Belcher et al. (2020) [23]				Distress: General stress, cancer-specific stress, cancer worry, depression, anxiety	Inability to make ends meet, not enough money for necessities, cutbacks and adjustments			Functional health: physical function; symptom burden; symptom interference; comorbidities; social role, ability

Table 2 (continued)

References	Direct costs	Indirect costs	Material resources	Psychological response	Financial coping behavior	Financial outcome	Health outcome	Risk factors
Newton et al. (2020) [31]	Monetary measures of financial toxicity: direct medical costs, direct non-medical costs	Monetary measures of financial toxicity: indirect costs		Subjective measures of financial toxicity: Distress	Objective measures of financial toxicity: Coping mechanisms	Subjective measures of financial toxicity: Indebtedness	Subjective measures of financial toxicity: Health outcomes	Treatment seeking behavior: Diagnosis, health literacy, self-advocacy, personal financial circumstances, perception of the health system, clinician's recommendations, private health insurance status, proximity to treatment centers
Tucker-Seeley and Thorpe (2019) [43]			Material domain: Material hardship, making ends meet, material disadvantages	Psychosocial domain: Financial stress, financial worry, financial satisfaction	Behavioral domain: Financial adjustments, financial planning, spending/consumption			
Santacroce and Kneipp (2019) [30]	Financial costs: Direct costs	Financial costs: Indirect costs		Financial distress: Biological response to stress	Financial distress: Financial coping behavior	Family financial outcomes: Recent material hardship and impoverishment	Parent health outcomes: Symptoms, quality of life, risk for disease onset; Child health outcomes: symptoms, quality of life, relapse	Pre-existing parent factors: age, gender, language, marital status, work status; Pre-existing family factors: residence, income, assets, debts, number of children, general health; Post-cancer diagnosis: treatment decision and initiation
Carrera et al. (2018) [32]	Objective financial burden: Expenditures on drug costs, other direct medical costs, treatment costs		Subjective financial distress: Wealth – wages, salaries or replacement income; savings and assets	Subjective financial distress: anxiety and discomfort				

Table 2 (continued)

References	Direct costs	Indirect costs	Material resources	Psychological response	Financial coping behavior	Financial outcome	Health outcome	Risk factors
PDQ Adult Treatment Editorial Board (2002) [5]	Medical costs, non-medical costs			Financial strain and distress		Formal bankruptcy	Health outcomes	Pre-illness health, assets, debt, income, illness or injury, medical insurance, treatment choice
Pisu et al. (2010) [25]	Direct costs: medical costs, non-medical costs, time costs	Time costs (at work) Indirect costs: Productivity losses due to cancer					Psychosocial cost: loss of quality of life	
McNulty and Khera (2015) [45]				Patient/family: Increased stress and uncertainty	Spending savings/retirement funds, borrowing money, losing home/selling property, lifestyle changes, avoiding purchases, reduced spending on food and clothing, changes in decision making/priority selling; Cancer treatment: decreased adherence to cancer treatment	Incurring significant debt, bankruptcy	Patient/family: Parent cost, disability Decreased health related quality of life: Increased stress, uncertainty, anxiety and depression, demands on the caregiver	Risk factors: Patient/family socio-demographics, financial/employment, environmental/logistical, cancer and disease related
Lentz et al. (2019) [24]	Costs related to medical, surgical and radiation treatment, including costs of supportive care, end-of-life care	Material consequences: Reduced income		Psychological consequences: Distress	Maladaptive coping: Skipping or reducing medication doses, adjusting non-medical spending	Material consequences: Depletion of savings, debt, bankruptcy	Psychological consequences: reduced quality of life	Causes: Baseline factors: demographics, health, socioeconomic status Cancer: type and stage Medical insurance status: Premiums, deductibles, coinsurance, absence of

OOP out of pocket

Table 3 Number of a priori (sub)concepts in each model

Concept Sub-concept	Model Reference No.																		Total
	[28]	[26]	[2]	[3]	[30]	[33]	[34]	[27]	[42]	[23]	[31]	[43]	[30]	[32]	[5]	[25]	[45]	[24]	
Direct costs	x	x	x	x	x	x		x	x		x		x	x	x	x		x	14
Direct medical costs	x	x						x	x		x			x	x	x		x	9
Direct non-medical costs		x						x			x				x	x			5
Indirect costs		x		x	x	x			x		x		x			x		x	9
Income loss		x		x		x			x									x	5
Time loss at work		x		x							x					x			4
Material resources			x	x		x			x			x		x					6
Financial coping behavior		x	x	x	x	x		x		x	x	x	x				x	x	12
Increase in resources		x															x	x	3
Labor substitution		x																	1
Reduction in expenditure		x															x	x	3
Treatment adherence			x	x				x			x						x	x	6
Financial outcome											x		x		x		x	x	5
Health outcome	x										x		x		x	x	x		6
Psychological response	x		x	x	x	x		x	x	x	x	x	x	x	x		x	x	15
Cognitive response	x		x	x					x	x		x							6
Physiological response			x	x		x		x		x	x	x	x	x			x	x	11
Risk factors		x					x	x	x	x	x		x		x		x	x	10
Disease characteristics		x								x			x		x		x	x	6
Employer factors							x												1
Environmental factors																	x		1
Health care system factors							x												1
Health insurance								x							x			x	3
Health Provider factors							x												1
Household factors							x						x				x		3
Individual factors		x							x	x	x				x		x	x	7
National level factors							x												1

x indicates the presence of the (sub)concept

> The medical and non-medical costs associated with the diagnosis of childhood leukaemia … (Quote from authors [36])
> Some costs were directly related to treatment/care… Some other costs were more incidental and related to the physical, psychological and social effects of cancer… Increased domestic fuel bills were common… nutritional supplements, complementary therapies, wigs… (Quote from authors [35])
> The cost of being in hospital was quite high, for myself and my family because they were visiting every day, paying car parking (Quote from patients [35])

'Indirect costs' were also mentioned consistently [35, 37–41], including 'time loss at work':

> …most patients did not continue working normally during their illness and treatment…some were able to work reduced hours, or at home… (Quote from authors [37])
> …I just went back…probably about 10 months later I was made redundant. (Quote from patients [39])

and 'income loss':

> All the patients who stopped working after their diagnosis experienced a drop in income. (Quote from authors [37])

Definitions of direct and indirect costs appeared in three models [26, 31, 42]. Two models [26, 31] described them in terms of their sub-concepts, one model [42] provided a specific definition in the context of prostate cancer. As a result, we developed a general definition for 'direct costs' and 'indirect costs', provided in Table 5. The definitions of related sub-themes under the themes of direct and indirect costs in the model of Kankeu et al. [26] were used verbatim in our definitions (see Table 5).

3.3.1.2 Position in our conceptual framework Compared with other included themes, prior models showed direct

Table 4 Definitions of (sub)concepts derived from existing models

Concepts	Definition	Sub-concepts	Definition	References
Direct costs (OOP expenditure)	**Including direct medical and non-medical costs due to cancer**	Direct medical costs	Financial costs of health care (consultation, medicines, laboratory, hospitalization, etc.)	[26, 31]
		Direct non-medical costs	Other financial costs related to seeking care (transportation, special dietary regimens, etc.); costs to manage complications and/or other side effects; time costs of patients, time costs of caregivers	
Indirect costs (productivity loss)	**Loss of working time of person who is ill and caregivers due to cancer; loss of income of person who is ill and caregivers (due to absenteeism, missing business appointments, etc.) due to cancer; any lost salary from premature mortality due to cancer can also be considered**	Time loss at work	Loss of working time of person who is ill and of caregivers	[26, 42]
		Income loss	Loss of income of person who is ill and caregivers (due to absenteeism, missing business appointments, etc.); any lost salary from premature mortality can also be considered	
Material resources	**Total OOP expenditure as percentage of household income, available savings and assets**			[2, 32, 42]
Psychological response	Psychological response to the increase in household expenses that must now be managed as patients navigate cancer care	**Cognitive and emotional response**	**Concern about wages/income meeting expenses related to costs of cancer care**	[3, 30]
		Physiological stress	**Biological responses to stress**	
Financial coping behavior	The behavior that patients and their households adopt to the cost increase due to cancer	**Increase in resources**	Borrowing money; using savings; selling assets	[3, 24, 26, 45]
		Reduction in expenditure	Reducing/delaying consumption of non-health goods and services (food, education, electricity, leisure, etc.); delaying investments	
		Treatment adherence	Took less or skipped medication/treatment; delayed or missed physician visit	
		Labor substitution	**Intra-household labor substitution; hiring other labor and other strategies**	
Financial outcome	**Incurring significant debt, bankruptcy or poverty**			[5, 24, 30, 31, 45]
Health outcome	**Anxiety, depression, reduced quality of life and worsening health status (on patients and family members)**			[28, 30, 45]

Table 4 (continued)

Concepts	Definition	Sub-concepts	Definition	References
Risk factors		Disease characteristics	Cancer type, stage, treatment, side effects…	[24, 30, 45]
		Individual factors	Baseline factors: demographics, health status, socioeconomic status…; patient-specific values: a patient's personal preferences and perceptions, which influence how they react to medical-related expenses and are the most difficult contributors to generalize, may also be related to treatment choices and health insurance selection; self-management behaviors: health directed activity, positivity, active engagement in life, self-monitoring and insight, constructive attitudes and approaches, skill technique and acquisition, social integration and support, health service navigation	[23, 24, 42]
		Household factors	Demographics, member's health, socioeconomic status, household size, work status…	[30, 34, 45]
		Employer factors	Health insurance coverage, paid and unpaid sick leave, workplace accommodations	[34]
		Health provider factors	Health provider (doctors, nurses...): age, year of medical school graduation, training and specialty, geographic region and practice setting	[34]
		Health care system factors	Breadth and depth of provider networks, benefit design, routine use of electronic health record functionality in quality improvement and financial assistance infrastructure	[34]
		National level factors	How do state-level policies, such as Medicaid eligibility threshold and expansion status, affect financial hardship? How do policies related to generic substitution, cost transparency and oral parity affect financial hardship? Do policies related to availability of marketplace coverage, essential health benefits, or elimination of pre-existing coverage exclusions affect hardship?	[34]
		Geographical factors	Having to relocate during treatment; longer distance from hospitals/cancer centers; rural dwellers	[34]

In bold: (Sub)concepts and definitions that will be changed during the synthesis of qualitative studies. Compare with Table 5 for specific changes

OOP out of pocket

Table 5 Definitions of (sub)themes in the Socioeconomic Impact Framework

Themes	Definition	Sub-themes	Definition
Direct costs (OOP expenditure)	Costs due to cancer paid by patients and/or their households (out-of-pocket costs), including direct medical and non-medical costs	Direct medical costs	Financial costs of health care (consultation, medicines, laboratory, hospitalization, etc.)
		Direct non-medical costs	Financial costs related to seeking care (transportation, special dietary regimens, etc.); cost to manage complications and/or other side effects
Indirect costs (productivity loss)	Costs indirectly incurred from cancer. From patients and household perspectives, it is assumed as productivity loss, including time loss at work and income loss	Time loss at work	Loss of working time of person who is ill and of caregivers
		Income loss	Loss of income of person who is ill and of caregivers (due to absenteeism, missing business appointments, etc.) Any lost salary from premature mortality can also be considered
Financial coping ability	Patients' and their households' financial ability to cope with the cost of cancer care	Household health expenditure ratio	Total OOP expenditure as percentage of household income
		Household available savings and assets	Available savings and assets of patients and their household
Psychological financial response	Psychological perception of the increase in household expenses that must now be managed as patients navigate cancer care	Financial experience	Perception (feeling) towards the financial experience to meet expenses related to costs of cancer care (past and present). This can be negative financial rumination and/or positive satisfactory perceptions
		Financial expectation	Perception (feeling) towards the future expectation of financial ability to meet expenses related to costs of cancer care. This can be negative worry thinking about the ability to work, or possibility having to use savings, assets… and/or positive motivation to control the future financial situation (extra work, financial coping behavior…)
Financial coping behavior	The behavior that patients and their households adopt in response to the cost increase due to cancer	Increase in liquidity and resources	Behavior such as borrowing money; use of savings; sale of assets; extra work or early return to work to increase household income; requesting social benefits/support…
		Reduce in expenditures	Behavior such as reducing/delaying consumption of non-health goods and services (food, education, electricity, leisure, etc.); delaying investments
		Treatment adherence/changes	Taking less or skipping medication/treatment; delaying or missing physician visits

Table 5 (continued)

Themes	Definition	Sub-themes	Definition
Risk factors	Factors associated with an increased risk of socioeconomic impact of cancer	Disease characteristics	Cancer type, stage, treatment, side effects…
		Individual and household factors	Baseline factors: demographics, patient's and member's health status, socioeconomic status, household size, existing mortgage, number of dependents… Individual-specific values: a household member's personal preferences, self-efficacy. This may also relate to treatment choices, health insurance selection Self-management behaviors: health-directed activity; positive, active engagement in life; self-monitoring and insight; constructive attitudes and approaches; skill technique and acquisition; social integration and support; health service navigation Household relationship: family roles, communication
		Societal factors	Financial support and emotional support from different levels of society: friends, employer, health provider e.g., financial support: informal financial support, health insurance scheme, social benefits, employer flexibility… Emotional support: Social stigma, social relationship, emotional support from peers, health professional…
		Contextual factors	Contextual factors: health care system, national system and policies

OOP out of pocket

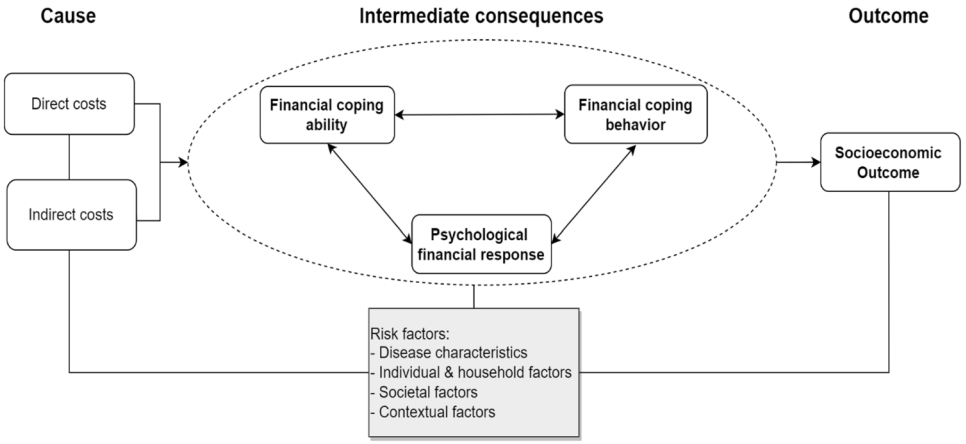

Fig. 3 Socioeconomic impact framework

and indirect costs in parallel on top of a hierarchy-like model [33] or at the primary points (normally on the left side) of many process-like models [26, 27, 30, 31]. As a result, direct and indirect costs, in parallel, appeared as the causes in our conceptual framework (see Fig. 3).

3.3.2 The Intermediate Consequences—the Dynamic Loop of 'Financial Coping Ability', 'Psychological Financial Responses' and 'Financial Coping Behavior'

3.3.2.1 Adjustment from the a priori concepts Substantial changes were made to create the dynamic loop of three critical themes in our conceptual framework, including 'financial coping ability', 'psychological financial responses' and 'financial coping behavior'.

First, we changed the concept 'psychological response' to the theme 'psychological financial responses' to stress the financial impact of cancer and to distinguish this from other psychological responses at the cancer diagnosis stage that were mentioned in some qualitative studies.

> …there was a remarkable variation in the incidence of parental psychological dysfunction that resulted from the disclosure of the medical diagnosis (Quote from authors [36])
> …their shock and fear on receiving cancer diagnosis (Quote from authors [39])

We use the concept, 'psychological financial responses' to signify psychological effect as a reaction to the costs of cancer during the course of disease, as also indicated in some of the qualitative studies.

> …financial issues associated with cancer could provoke a variety of negative emotions such as regret, disappointment and self-reproach. (Quote from authors [35])
> "I am getting depressed. So I go. The house is going up for sale" – The wife is going berserk because she cannot keep up the bills in the house (Quote from patient's family member and authors [35])
> Some patients who had stopped working during treatment were concerned about managing financially on sick pay and/or benefits. (Quote from authors [37])

The renaming of 'psychological responses' to 'psychological financial responses' did not affect its definition, taken verbatim from Altice et al. [3] (see Table 5). We also decided to exclude the a priori sub-concept of biological responses as we believed that this sub-concept is more related to health-related impacts rather than the SEI of cancer.

Additionally, some qualitative studies [35, 38] found respondents worried not only about their current financial situation but also about their financial future.

> Some participants felt that their financial hardship and/or reduced lifestyle would last for the foreseeable future. (Quote from authors [35])

Another study reported stress about future savings and retirement.

> As well as being worried about their current situation, patients described worries about the future. These worries include: not being able to replenish savings used during illness…implication of using money saved for retirement… (Quote from authors [38])

One participant reported concern about future ability to work.

> The worry of my pension and of not knowing when I am going back to work… (Quote from patient [35])

It is not to be assumed that all (or most) patients experience financial difficulty or any psychological distress resulting from it. Some researchers found that not all patients who

experienced changes in their financial situation are necessarily burdened by it.

> …A breast cancer patient received full sick pay, had private health insurance, and obtained a medical card post-diagnosis… A retired married prostate cancer patient had surgery, few side-effects, private health insurance, was eligible for a medical card…Neither of these patients reported any financial distress (Quote from authors [38])

Therefore, under the theme 'psychological financial responses', we excluded the a priori sub-category 'cognitive response (worries)' and replaced it with the more neutral sub-themes 'financial experience' and 'financial expectation'. They are listed along with their definitions in Table 5.

Moreover, several of the qualitative studies [37–39] illustrated that the material resources themselves were less important to the experience of SEI of cancer than how far the assets and liabilities of individuals gave them the ability to financially cope. We therefore changed the a priori concept 'material resources' to the theme 'financial coping ability'. The term 'ability' was mentioned by Moffatt and Noble [39].

> The sudden drop in income experienced by many of our participants affected their ability to meet bills, housing payments and other bills. (Quote from authors [39])

We established two sub-themes under the theme 'financial coping ability', including 'household health expenditure ratio' and 'available household savings and assets'. The sub-theme 'household health expenditure ratio' was mentioned as the total OOP expenditure as percentage of household income in one included model [2]. Available savings and assets were also mentioned in the a priori concept 'material resources' and included in models [2, 32, 43]. We added the term 'household' to clarify the focus on household unit of this sub-theme.

In the dynamic loop, the only a priori concept that remained unchanged was 'financial coping behavior', since it reflected the actual behavior of the patients and/or their family members dealing with the costs of cancer, and it appeared in all seven qualitative studies [35–41]. However, a minor change was made to its sub-concepts, in which we decided to merge the 'labor substitution' and 'increase of resources' sub-concepts together, since the behavior mentioned in 'labor substitution' also led to the increase of resources.

All of the changes can be observed when comparing Tables 4 and 5.

3.3.2.2 Position in our conceptual framework The relation between the causes (direct and indirect costs) and the dynamic loop: In general, the causes in our conceptual framework, 'direct costs' and 'indirect costs', are correlated with each theme in the dynamic loop (see Fig. 3). Several studies [35, 38–40] mentioned the difficulties that individuals experienced in meeting their expenses as a result of increased costs and income loss. This dynamic is represented in the relationship between the causes and three themes in the loop.

> The sudden drop in income experienced by many of our participants affected their ability to meet bills, housing payments and other bills. (Quote from authors [39])
>
> Potential major changes in the lifestyle of the family to meet the demands of the new situation were also among the factors found to exacerbate the parental sense of lack of control (Quote from authors [36])

Additionally, existing models established a relationship between direct costs, indirect costs and 'psychological financial response' [5, 28, 30, 33]. Similarly, the causes (direct and indirect costs) also lead to various 'financial coping behaviors', as shown in previous models [24, 26, 31, 33].

3.3.3 The Relationship amongst Themes in the Dynamic Loop

The three themes ('financial coping ability', 'psychological financial response' and 'financial coping behavior') are also tightly linked to each other. In one model [29], the authors included a mediator between 'financial psychological response' and depleted savings as well as an increase in debts, which is closely related to the theme 'financial coping behavior'. In the reverse direction, the behavior also caused a more severe psychological response. Evidence from one qualitative study [38] supported both directions.

> …patients who were worried about being able to afford to buy medications or considering stopping treatment because they could not afford it. (Quote from authors [38])
>
> …patients were generally uncomfortable with asking for, or accepting help, particularly from charities, and this caused stress and worry. (Quote from authors [38])

Carrera et al. [32] pointed to a bi-directional relationship between 'wealth' and 'anxiety and discomfort'. While anxiety and discomfort could be regarded as the 'financial psychological responses', wealth can be related to the 'financial coping ability' themes. This bi-directional relationship was also supported in the results of two other qualitative studies.

> Pre-existing routine non-cancer related expenses (such as mortgages) could become more onerous as household income reduce. Because of these…most

> caregivers experienced considerable concern about their financial situation (financial strain) … (Quote from authors [40])
>
> As well as worried about their current situation, patients described worry about the future...worry about: not being able to replenish savings used during treatment/illness… (Quote from authors [38])

In addition, 'financial coping behavior' is also closely linked to 'financial coping ability' themes, as is shown in the work of Tucker-Seeley and Thorpe (2019) [43] as a bi-directional relationship. There were also observations by authors from another study [37] that further illustrated this relationship.

> …a patient who had postponed treatment until she received a medical card. A number of patients described how stressful hospital bills were while they are waiting for a medical card. (Quote from authors [37])
>
> Patients reported struggling financially. Difficulties managing the weekly household budget were common. All patients…have to budget and spend more carefully following diagnosis. (Quote from authors [37])

In conclusion to this dynamic, we propose a loop that involves three themes: 'financial coping ability', 'psychological financial responses' and 'financial coping behavior', as intermediate consequences resulting from the financial changes due to cancer care. These components are closely linked by bi-directional relationships among one another and are caused by direct and indirect costs of cancer care.

3.3.4 The Theme 'Risk Factors'

3.3.4.1 Adjustment from the a priori concepts Categorization of the a priori concept of risk factors was taken from Yabroff et al. [34]. In this model, the authors defined the risk factors by providing a list of examples under each category. Since our conceptual framework focuses on the patient and household, we decided to recategorize the a priori sub-concepts of 'risk factors' to form an overarching theme of 'risk factors' in our framework. First, we kept the sub-concept of 'disease characteristics' as it was to be a sub-theme in our conceptual framework. Second, we merged 'individual and household factors' into one sub-theme. Then, we redefined the 'societal factors' sub-theme by grouping the sub-concepts 'employer factors' and 'health provider factors'. Lastly, the sub-theme 'contextual factors' were formed by grouping sub-concepts 'health care system' and 'national level factors'.

After these adjustments, we defined the theme 'risk factors' as "the factors associated with an increased risk of SEI of cancer" (see Table 5).

3.3.4.2 Position in our conceptual framework In over half of the included qualitative studies [36–39], various risk factors were mentioned, for example:

> …various possible risk factors for financial difficulties: working at diagnosis; having young children; being a lone parent…lack of social/family support (Quote from authors [37])

It is impractical to list all of the risk factors, rather we can include some examples in our definitions. We proposed potential links between the theme 'risk factors' with all other themes in our conceptual framework (see Fig. 3). The exact risk factors and their links to other themes falls outside the scope of our conceptual framework.

3.3.5 The Proposed 'Socioeconomic Outcome of Cancer'—a Multi-dimensional Outcome

First, we decided not to include the 'health outcome' concept as a theme in our conceptual framework as it is well established in previous research [44]. Instead, we focus only on the financial-related impact of cancer. Second, the concept 'financial outcome' referred to debt, bankruptcy or poverty in existing models [24, 30, 31, 45]. This certainly can be a consequence of the 'financial coping behavior' of patients and their household. However, we also observed other outcomes, for example, the worry of patients (or their household member) on how to deal with the changes in finances, which we described as part of the 'psychological financial response' theme (see Table 5).

Consequently, we propose a broader concept to cover all the consequences from the changes in finances in cancer care among patients and their households, called the 'socioeconomic outcome of cancer'. This outcome should be understood as a multi-dimensional outcome that results from the intermediate consequences as described in the dynamic loop above, including 'financial coping ability', 'financial psychological responses' and 'financial coping behavior'. We suggested the term of 'socioeconomic outcome of cancer' for this outcome and included it at the end point of our proposed conceptual framework (see Fig. 3).

In Fig. 4, we integrated our Socioeconomic Impact Framework with the health-related quality of life (HRQoL) model by Valderas and Alonso [44] to support our hypothesis of their linkage to contribute to the concept of overall quality of life. In their health-focused model, different constructs are linked with each other to deliver the outcome of HRQoL. We propose that various constructs in our conceptual framework contribute to what we call socioeconomic outcome. Both of these components, potentially among others, contribute to the overall quality of life of an individual, as shown in the combined model in Fig. 4.

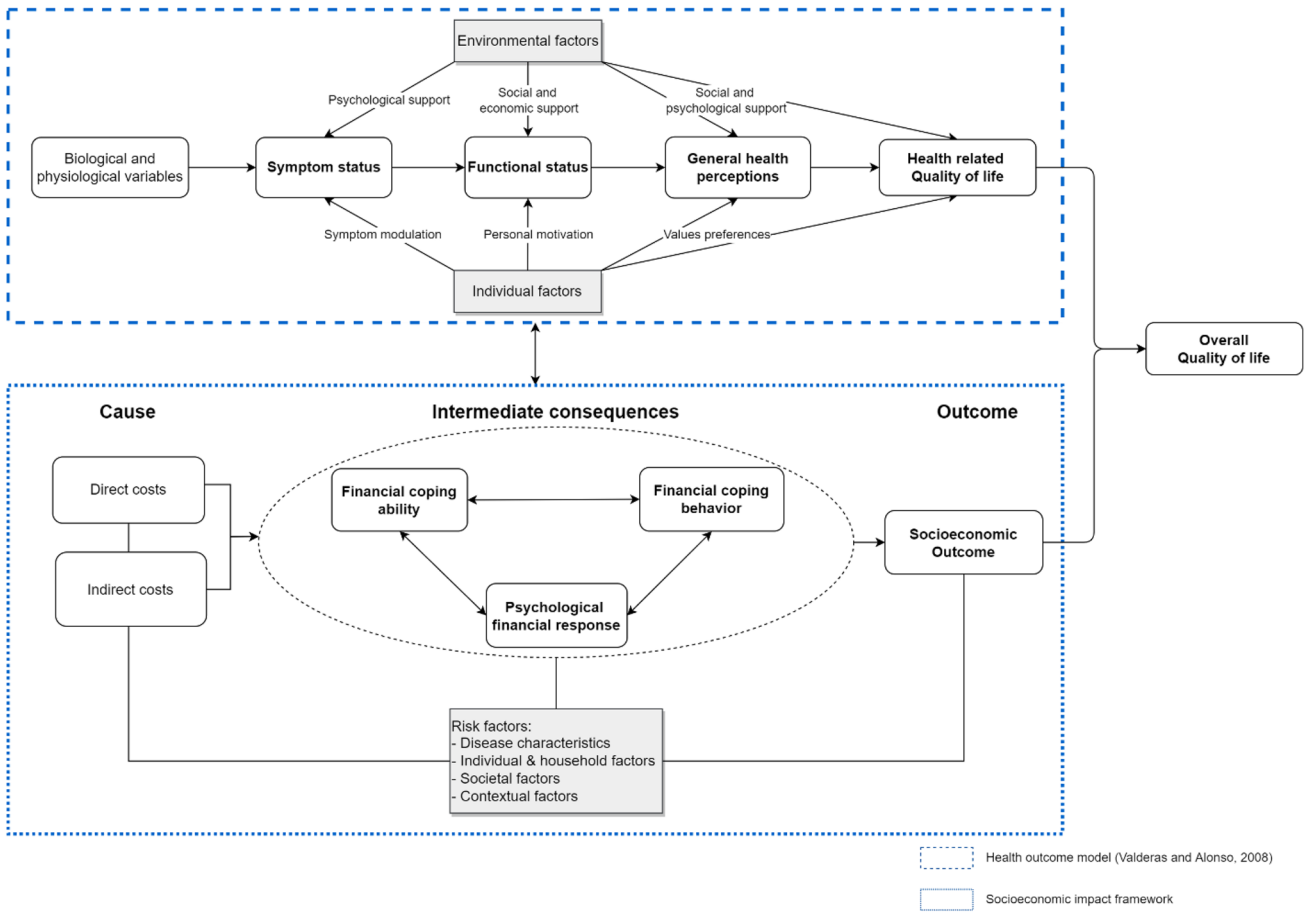

Fig. 4 An integrated framework: Health outcome model and Socioeconomic Impact Framework

3.4 Sensitivity Analysis Results

The exclusion of five US-based models [3, 5, 23–25] and one low- to middle-income country-based model [26] had a negligible impact on the number of a priori (sub)concepts (Supplementary Material 2.4, see ESM). Only one subtheme (labor substitution) was omitted as it only appeared in a low- to middle-income country-based model, which was excluded in sensitivity analysis [26].

Supplementary Material 2.5 presents the definitions of a priori (sub)concepts that were derived from existing models, both before and after excluding US-based and low- to middle-income country-based models (see ESM). Many of the definitions remained unchanged, including concepts like 'direct costs,' 'financial coping behavior,' 'financial outcome,' as well as sub-concepts like 'physiological stress,' 'increase in resources' and 'reduction in expenditures.' Some definitions were reworded because of the exclusion of US-based and low- to middle-income country-based models, but the overall meanings were not significantly altered. For example, the 'direct medical costs,' 'direct non-medical costs' and 'treatment adherence' sub-concepts underwent slight wording changes. For (sub)concepts, more detail was added when considering all existing models instead of excluding the US-based and low- to middle-income country-based ones. These included concepts like 'indirect costs,' 'health outcome' and sub-concepts like 'time loss at work,' 'income loss' and 'individual factors.' It is also worth noting that in some cases, the definitions derived from all models were more general compared with those derived from the sensitivity analysis. For example, the definitions of the 'indirect costs' concept or 'disease characteristics' sub-concepts focused on one particular cancer type if excluding US-based and low-middle-income country-based models, while their definitions referred to all cancer types without the exclusion (see Supplementary Material 2.5 in the ESM).

4 Discussion

4.1 Main Findings

This study had the objective to review available concepts and models in the research area of SEI of cancer and adapt the

suitable concepts to the European context. As a result, we propose a comprehensive Socioeconomic Impact Framework (Fig. 3) that presents seven themes and their relationships in relation to the SEI analysis of cancer, from the perspectives of patients and their households. Under the seven themes, there are 15 sub-themes included. We suggested a clear definition for each theme and sub-theme (see Table 5) based on the collection of previous definitions used in existing models in combination with evidence from qualitative studies in the European context.

4.2 What is New About Our Proposed Conceptual Framework?

To our knowledge, this is the first study that performed a targeted review and framework synthesis of available conceptual models in the field of interest, the SEI of cancer. We propose this broad concept of SEI to cover various terms used in the included models, for example, financial burden [28, 31, 36, 37], financial stress/strain [23, 28], financial distress [24, 37], economic/financial hardship [3, 28, 45]. Our review has an advantage of identifying the commonalities and differences among available concepts and their definitions in existing models, and we suggest more general terms and definitions. We are in the process conducting a more in-depth analysis of terminology used in studies conducted in Europe with the aim of recommending a consistent terminology use in this field of research in Europe.

Second, this is also the first study that combines the breadth of existing models with evidence from qualitative studies using thematic analysis to create an adapted framework focusing on the European context. This was accomplished by applying a best-fit framework synthesis. None of the previous models focused on the European context (see Supplementary Material 2.2 in the ESM). Five models specify the US as the region of interest [3, 5, 23–25], one model focuses on Australia [31], one model targets low- and middle-income countries [26], while the eleven remaining models did not specify the geographical context. Given the differences in health systems among countries [46–48], we believe that a context-specific framework could generate more meaningful information for policy making. This line of thinking is akin to the realist approach, which is a theoretical- and contextual-driven evaluation approach for evidence-based policy [49]. Furthermore, this approach is getting more attention in the field of implementation science and health economics in recent years [50, 51].

Finally, we propose that the socioeconomic outcome of cancer is supplemental to HRQoL; these two outcomes combined contribute to the overall quality of life (see Fig. 4). Our rationale is as follows: first, the SEI of cancer has been shown to relate to HRQoL [52–54]. Second, HRQoL focuses on the health-related aspects of an individual, which means that elements beyond health are less precisely conceptualized. Therefore, we tend to support the latter notion of considering 'socioeconomic outcome' as a supplemental but distinct component to HRQoL, which together contribute to the overall quality of life of an individual.

4.3 Strengths and Limitations

There are three strengths of the methodology, which were also mentioned in the original papers of best-fit framework analysis [17, 18], used in this study. First, the systematic inclusion of existing models for framework synthesis helped to overcome the limitations of one single model. As aforementioned, the a priori concepts in included models cover all available concepts in a consistent manner. Second, this methodology employs a unique combination of framework synthesis and thematic analysis to adapt the existing conceptual backgrounds into a specific context of interest.

The sensitivity analysis we conducted revealed that excluding models focused on the US population and low- to middle-income countries did not significantly impact the number of included a priori (sub)concepts. Only one subtheme 'labor substitution' was omitted as it only appeared in a low- to middle-income country-based model [26]. This change is consistent with the adaptation in our conceptual framework after considering the 'labor substitution' behavior as part of the 'increase in resources' sub-theme. It is important to note that no particular model focused on the European context. Therefore, evidence from European qualitative studies provided a valuable contribution to our conceptual framework. The inclusion of this evidence ensures that our framework was comprehensive and relevant to the European context. Indeed, this would be valuable not only for researchers in the field, but also for policy and decision makers. For example, healthcare professionals may recommend that patients look for social benefits if they see that the patients are facing financial problems. Policy makers also could think more thoughtfully on how to better structure the social benefit scheme if taking the SEI into consideration. Third, another strength is the coherence in the thematic analysis process. Coding against an a priori framework prevents redundancy among the coders and helps to generate the comprehensive discussion to facilitate final agreement.

There are some limitations to this study. First, some relevant articles may be missed due to the use of only PubMed, EconLit and Web of Science as databases. Other relevant databases were not utilized in the review (e.g. Cochrane Central, Embase or CINAHL). Nevertheless, it is a limitation of any systematic review and we have tried to minimize it by cross-checking the articles identified in systematic literature reviews that searched for similar articles in addition to the initial electronic search process (see Supplementary Material 1 in the ESM). Second, the variety of terms and

definitions used in existing models generated difficulties for creating the a priori (sub)concepts. We had to find terms to generalize similar concepts with sometimes conflicting definitions. Although the process involved a discussion among team members to ensure that different perceptions of how these models can be combined are included, it is, nevertheless, a subjective process, which means the a priori concepts could also be differently structured. Another limitation is that our synthesis on qualitative studies was limited to the breadth of the results sections of these studies. This approach can capture the conceptualization from the authors by categorizing the participants' quotes into themes, but may not reflect all of the experiences of participants' that are relevant for this paper. In addition, as no existing model specifically focused on the European context was identified (see Supplementary Material 2.2 in the ESM), and the number of included qualitative studies was limited to only seven, there is a possibility that other relevant concepts that are specific to the European context may not have been covered.

The conceptual framework presented in our study is intended to provide an important contribution to a comprehensive European consensus project on the SEI of cancer. This project is led by a dedicated Task Force under the auspices of the Organization of European Cancer Institutes (OECI) [16]. Members are from various disciplines including clinical oncology, health economics, cancer research and patient advocacy, all with a European background. Given the expertise of the Task Force is rooted in Europe, we decided to stress the European context in the paper. For the same reason, we are cautious in recommending the framework be applied beyond the European context, while acknowledging that elements may be applicable in other regions. Moreover, the evidence for the relationships between themes and subthemes in our framework is based on qualitative analysis. As such, they can be understood as hypotheses for further research. Finally, other potential dimensions may not be covered by our proposed framework. One particular example of such a dimension that has been included in models is the time course of disease (e.g. diagnosis, treatment, disease-free survival) [26, 31]. These models explicitly cover the disease course after diagnosis. We, however, believe that each (sub)theme in our conceptual framework is relevant at any time of measurement; therefore, we decided against the inclusion of a temporal dimension.

5 Conclusion

We proposed the Socioeconomic Impact Framework by reviewing and synthesizing existing models and qualitative studies in the European context. The potential users of our proposed framework include not only researchers in the field, but also relevant decision makers in the European context. Primarily, since this study is conducted by a small group of authors with a background in health economics, our next aim is to acquire a consensus agreement on the terminology, definitions and relationships between the themes and sub-themes included in the framework for consistent use in future research in this area. Hence, our work contributes to a European consensus project on SEI research by an Organization European Cancer Institute (OECI) Task Force to consolidate the conceptual framework proposed in this study by acquiring reviews from European experts in the field [16]. As an outcome of this Task Force, a recommendation paper is expected to guide future research and to inform other researchers, healthcare providers, decision makers or other relevant stakeholders about the topic. Apart from this, further empirical research is necessary to investigate the overall concept of SEI as well as its relationship with HRQoL.

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Declarations

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Conflict of interest Phu Duy Pham, Jasper Ubels, Rachel Eckford, Karla Hernandez-Villafuerte and Michael Schlander have no competing interests to declare.

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Ethics approval The study was conducted on published literature, therefore ethics approval is not applicable.

Consent to participate Not applicable.

Consent for publication Not applicable.

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Author contributions The research was initiated and designed by PDP. PDP and JU conducted the analysis and interpretation of data. All authors contributed to the revision of the manuscript and approved its contents.

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