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RESEARCH

# Health-related roles of older generations in families with inherited genetic conditions: a scoping review

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## Abstract

Inherited genetic conditions are family diseases. They affect consanguineous relatives, in lineage for several generations, and impact the family dynamics. Older generations have been considered highly influential in the health management of families with inherited genetic conditions. To our knowledge, no reviews so far addressed the health-related roles of older generations in these families. This scoping review aims to fill that gap by mapping the existent research about the health-roles roles performed by the older generations in families living with autosomal dominant inherited genetic conditions. Four electronic databases were searched: Scopus, Web of Science, PubMed, PsycInfo. Eleven studies were included, and relevant findings were extracted. Main roles included: informers vs. blockers of disease-related information; encouragers vs. discouragers of health screening or genetic testing; (non-)supporters; and role models in living and coping with the disease. The roles played by older generations are relevant to the health management of other family members and can be beneficial to themselves (reciprocal interactions). Acknowledging and understanding these roles is important for professionals and health-services. Results suggest the relevance of an intergenerational perspective when working with families with inherited genetic conditions.

**Keywords** Older generations · Health-related roles · Family · Hereditary disease · Genetic disease

## Introduction

Inherited genetic conditions (IGCs) are family diseases. Like any other chronic condition, they impact the family dynamics and require family mobilization. Hereditary diseases are familial also in the sense that they affect relatives for several generations and therefore must be managed within the extended family system. Families must deal with the (risk of) transmission of the pathogenic variant among their members, whose understanding of the biological mechanisms are widely studied and rapidly evolving (Mendes et al. 2018).

Generational transmissions in families living with IGCs involve familial and relational processes that go beyond biological processes, which have been less explored (Seidi et al. 2023). Literature has mainly underlined family history (i.e. information about disease and risk, shared environmental and behavioral factors) (Claassen et al. 2010), family dynamics and adaptation to the IGC, health and risk management and life decisions, and the impact of genetic information in the family system (Sobel and Cowan 2000); and influence of the social context on the family (like guilt,

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secrecy or stigma) (McDaniel 2005; Mendes et al. 2017). Family processes are described as key to the health management of the condition, for example regarding adherence to preventive measures (Ashida et al. 2010; Saastamoinen et al. 2020), influencing communication and decisions about (not) undergoing genetic testing (Dimond et al. 2022; Law et al., 2022), treatments and/or reproductive decisions (Valdrez et al. 2014), as well as how individuals and families interact with their communities (Mendes et al. 2017).

Literature has been highlighting relevant roles of older generations who have been considered as "guardians of family memories" (i.e. those who strive for the preservation, conservation and transmission of the family identity among family members) (Vicente and Sousa 2010), "kin keepers" (i.e. those who are keen to stay updated regarding the medical/scientific issues of the condition, promote gatherings, and solidarity and connectedness among kin) (Troll and Bengtson 1992), or supporters (i.e. providers of emotional, practical and financial support) (Attar-Schwartz and Buchanan 2018; Ernsth-Bravell et al. 2016; Silverstein and Zhang 2020). Older family members have been considered influential in promoting healthy lifestyles among younger family members, including physical activity and smoking cessation (Escario and Wilkinson 2015; Palmer 2018). In families with IGCs, older generations are the most experienced in living with the condition in the family either because they carry it themselves or because they have been witnessing it in other family members over time. However, studies in families with IGCs focusing on the roles of older generations are limited. Previous reviews have mainly addressed communication in families with IGCs, including the communication between parents and their children and interactions among siblings (Atkinson et al. 2013; Chivers Seymour et al. 2010; Dattilo et al. 2021; Etchegary et al. 2013; Metcalfe et al. 2008; Shah and Daack-Hirsch 2018; Young et al. 2017).

To our knowledge, no reviews have been published addressing the health-related roles of older generations in families with IGCs. This scoping review aims to fill that gap by mapping the existent research on the health-roles roles performed by the older generations in families living with autosomal dominant IGCs. It also seeks to identify gaps in research evidence to guide future research that may contribute to improving the care that is provided to these families.

## Methods

This scoping study has been performed using the five stages of the framework adopted by Arksey and O'Malley (2005) and expanded by Levac et al. (2010) which included a systematic team approach throughout the entire process. Scoping reviews maps the literature and identify gaps in a particular research area (Arksey and O'Malley 2005). Data are reported following PRISMA 2020 (Page et al. 2021) (Fig. 1).

The research questions were defined according to the population, concept, and context (PCC) strategy: P, older generations in families with IGCs; C, health-related roles; C, extended family. This scoping review focuses on the health-related roles performed by the older generations. Therefore, the definition of older generations used for this review took into account that in families with severe late onset neurological diseases affected individuals will typically not reach old age due to shortened life-expectancy resulting from disease progression. Consequently, these individuals and families may have to deal with some developmental processes earlier in life than in non-affected families (Oliveira et al. 2022, 2023; Werner-Lin 2008). Therefore, older generation was defined considering the participants' generational position in relation to other family members (oldest family members) or their age (60 + years), independently of their genetic and kin status (carrier, non-carrier, at-risk, affected, nonbiological family member). Studies were excluded when the data did not consider the age or generation position between family members." Research questions were as follows: (i) What is known from the existing literature about the health-related roles performed by older generations in families with IGCs? (ii) What are the characteristics of older generations performing those roles (age, gender, and kinship)?

Inclusion criteria comprised original empirical qualitative, quantitative or mixed-methods studies published in peer-reviewed journals, written in English or Portuguese. The exclusion criteria were as follows: editorials, letters, commentary and opinion pieces, literature reviews, papers on theoretical issues, conference publications, books, unpublished materials such as thesis, dissertations, or abstracts; not addressing the health-related roles performed by older generations in families with IGCs.

Literature search strategies were developed using MeSH terms and text words related to older generations' health-related roles in families with IGCs. A preliminary search has been performed by the research team (with context expertise) to determine the relevant search terms, with the help of a librarian with methodological expertise in systematic/scoping studies search. The following terms were then determined related to the PCC:

["old* people" OR "old* person*" OR "old* adult*" OR aged OR elder* OR senior* OR "old* family member*" OR "old* relative*"] AND ["famil* communication" OR "health communication" OR "family network*" OR "famil* transmission*" OR "generation* transmission*" OR "intergenerational relation*" OR "social network*" OR "health

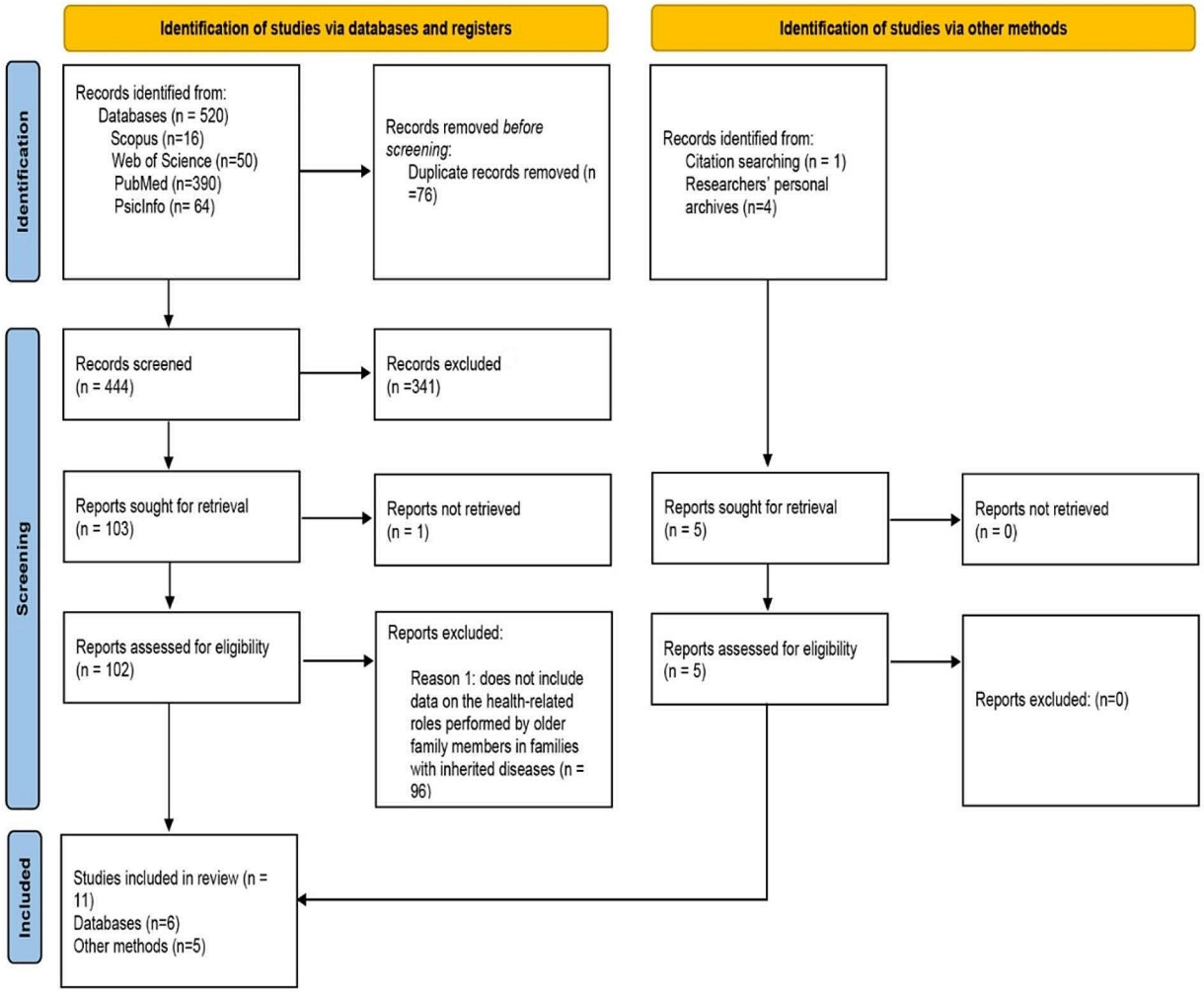


**Fig. 1** Flowchart of identification, selection, and inclusion of studies – PRISMA-ScR

promotion" OR "risk management" OR "health management" OR "social resources"] AND ["genetic disease*" OR "genetic condition*" OR "genetic illness*" OR "hereditary disease*" OR "hereditary condition*" OR "hereditary illness*" OR "inherited condition*" OR "inherited disease*" OR "genetic risk"].

Searches were performed in four Electronic Databases: Scopus, Web of Science, PubMed, PsycInfo. The electronic database search was supplemented by searches in the indexes of relevant journals, hand-search of reference lists and key-authors to identify additional relevant articles missed by this search strategy. Searches were conducted on 6th May 2021 with no restrictions on the date of publication.

After completing the searches, results were uploaded and managed using the reference manager software EndNote, including duplicates removal. After the duplicate's removal, the selection process (according to inclusion and exclusion criteria) was performed by two independent reviewers (CRO, CS) through two screening stages to make the final decision about the inclusion: (i) titles and abstracts and (ii) full articles. Both reviewers met at the beginning of the process to discuss any challenges or uncertainties considering the selection criteria. Papers were then independently double screened by each reviewer. Differences were discussed at the final of each stage until agreement on each paper was attained. Disagreements were resolved by the involvement of a third reviewer (ÁM). Reasons for papers exclusion were indicated at each screening stage (Fig. 1).

A total of 520 references were initially identified as potential papers for inclusion. 76 duplicates were removed. Based on titles and abstracts, 341 were excluded. The full text of 103 papers were read. After complete reading by the two reviewers, a total of six papers were selected for inclusion. Five additional papers were included from article

references and authors' personal libraries with a total of 11 studies included in the review (Fig. 1).

CRO and CS developed a data chart with the variables to extract according to the review questions. The following information was extracted: first author, year of publication, country, aims, methods, sample, IGC, older generations' roles, and implications. Relevant data from all studies were extracted and analyzed by CRO and ÁM (Tables 1 and 2). Next, an inductive narrative thematic approach was used for summarizing key findings across studies (Popay et al. 2006). The two researchers re-read through the extracted data and identified commonalities and differences that formed the basis for further synthesis. Based on commonalities and differences, the themes were generated through successive refinement. Discrepancies between researchers were resolved through discussion and consensus involving all the authors. Four main roles were identified: informers vs. blockers of disease-related information; encouragers vs.

**Table 1** Studies aims, methods and sample (chronological order)

| 1st author; year; country | Aims | Method | Sample |
|---|---|---|---|
| Koehly, 2009; USA | Identify the characteristics of gatherers, disseminators, and blockers of health information flow within BRCA1/2 mutation-positive families. | Quantitative, cross-sectional. | $N=183$ women from 124 families with known mutations in the BRCA1/2 genes; ages 22–57 years; average age: 40 years; 30% had a personal history of cancer; 85% were carriers of a BRCA1/2 mutation. |
| Ashida, 2010, USA | Evaluate associations between social network members who encourage screening (older, same, and younger generations) and individuals' motivation to undergo three types of screening: blood cholesterol, blood pressure, and blood glucose. | Quantitative cross-sectional. | $N=452$ adults; Mexican origin from 162 households; ages 20–75 years; average age: 43 years; 53.5% females; 24% at risk for heart disease; 33% at risk for diabetes. |
| Ashida; 2011; USA | Evaluate the role of older family members as providers of social resources within familial network systems affected by an inherited cancer susceptibility syndrome. | Quantitative cross-sectional. | $N=206$ from 33 families; people aged 18 to 83 years; average age: 43.8 years; 18.9% ≥ 60 years; 59.7% females; 27.9% with cancer history. Risk level: 24.8% have mutation; 34.5%; no mutation; 16% at risk (not tested); 24.7% not at risk. |
| Lehmann; 2011, UK | Views and experiences of grandmothers who have had carrier testing for one of two X-linked conditions: Duchenne muscular dystrophy (DMD) or Fragile X (FraX). | Qualitative. Thematic analysis. | $N=13$ grandmothers from families with Fragile X (8) and Duchenne muscular dystrophy (5); ages: 49–87 years; average age: 63.2; 10 have affected grandsons, 3 with daughters who chose not to continue with affected male pregnancies after prenatal diagnosis. |
| Mendes; 2012; Portugal | Explore families' experiences of cancer genetic counselling, through a familial perspective. | Qualitative exploratory. Grounded theory. Content analysis. | $N=9$ families (50 participants) (with at least 3 known mutation cancer carriers). 88% first-degree relatives (parents, offspring, and siblings); others were spouses. 5 affected by HNPCC, 3 by HBOC, 1 by HDGC mutation. Ages 15 to 78 years old. |
| Ashida 2015; USA | Evaluate psychosocial factors associated with sharing of family health history about heart disease and cancer among older adults. | Quantitative, cross-sectional. | $N=110$ older adults; ages 57–90 years; average age: 73.3; 78% females; diagnosis: 19.6% cancer, 26.2% heart disease. |
| Abad; 2017; Philipines | Explore genetic information communication in Filipino families affected with congenital adrenal hyperplasia (CAH). | Qualitative. Thematic analysis. | $N=5$ Filipino families with a child diagnosed with CAH (11 individuals). Ages: 18–62 years; average age: 44.5; all females. |
| Oliveira, 2017a; Portugal | Analyse the role of older family members (including carriers and non-carriers) towards younger ones, in terms of health promotion behaviours and genetic risk management, in families with paramyloidosis. | Qualitative exploratory. Content analysis. | $N=18$ members from 11 families with TTR-FAP; ages: 18 to 65 years; average age: 42; 10 women; 11 symptomatic carriers (8 with transplant, 3 medication), 2 pre-symptomatic carriers, 4 non carriers, 1 at risk. |
| Oliveira, 2017b; Portugal | Explore the intergenerational flow by analysing who from the older generation plays what roles (based on Oliveira et al. 2017a) towards whom from the younger generation. | Qualitative exploratory. Content analysis. | $N=18$ members from 11 families with TTR-FAP; ages: 18 to 65 years; average age: 42; 10 women; 11 symptomatic carriers (8 with transplant, 3 medication), 2 pre-symptomatic carriers, 4 non carriers, 1 at risk. |
| Pantaleao, 2019, USA | Identify key healthcare roles for managing LFS-related cancer risk and treatments held by parents and members of the younger generations in families living with LFS. | Qualitative study. Thematic Analysis. | $N=23$ families of 2–5 members (62 individuals); ages: 7 to 81 years; 56.5% females. |
| Oliveira; 2021; Portugal | Describe how adjustment to Huntington disease occurs, from a family perspective, considering the roles performed by older generations. | Qualitative exploratory. Thematic analysis. | $N=10$ members of 7 families with HD; ages: 28–72; mean age: 46; 8 females; 2 non-carriers; 2 pre-symptomatic carriers; 6 non-biological family members. |

**Table 2** Main findings of the included studies

| 1st author; year; country | Inherited genetic disease | Health-related roles of the older generation | Implications on family health-management |
|---|---|---|---|
| Koehly, 2009 USA | Hereditary breast ovarian cancers (HBOC) | Most frequently the older relatives are gatherers: searched for new information about cancer or genetic testing. Gatherers tend to be female and older first-degree relatives (parents) because they are often the gatekeepers to the health information of older and more distant family members for their children. Less frequently they are disseminators: spread genetic and cancer risk information to other family members and encouraged cancer risk discussions. Some are blockers: reluctance about learning or transmitting health information. | Older family members are a critical source of family health information. Gathering accurate family health history should engage the older generation. Older generations play an important role in answering children's questions regarding biology and genealogy.<br>Reciprocity: engaging older family members inherently enhances their health through improved social engagement within the family. |
| Ashida, 2010, USA | Cardiovascular disease | Screening encouragers: encourage screening and individuals' motivation to undergo three types of health screening: blood cholesterol, blood pressure, blood glucose. Mainly older first-degree relatives (parents): 28.3% mothers and 16.7% fathers; 6.8% of aunts and uncles and 6.9% of grandparents. | Older generation members encouraging screening is associated with higher levels of intention to screen.<br>Reciprocity: Involvement of older generations will allow them to play important social roles, increasing reciprocal interactions and enhanced life satisfaction. |
| Ashida, 2011, USA | Lynch syndrome | Screening encouragers. Communication about genetic counselling and testing and providers of health information. Providers of support: instrumental; help in crisis; emotional; dependability when needed; advice. Female were more likely to be listed as encouragers of colon cancer screening. | Benefits from targeting older family members as lay health advisors to facilitate communal coping processes and increase colonoscopy participation. Empower older individuals about their social roles in enhancing the well-being of their family members and to inform younger individuals about their older relatives' resourcefulness. |
| Lehmann, 2011. UK | Duchenne muscular dystrophy (DMD) or Fragile X (FraX). | In Fragile - X: seekers of information through professional, family and support group sources. In both conditions: making sense of the family pattern of the past (thinking about the way in which the gene had been inherited in the family; looking back in previous generations). Grandmothers: aged 49–87. | Addressing grandmaternal beliefs about the condition may be explored in the provision of psychosocial support. Involving the grandmother's partner should be encouraged. |
| Mendes 2012; Portugal | Hereditary cancers | Genetic counselling, testing, and screening encouragers of younger generations. Keepers of families' health history, described as valuable resources and as providers of privileged health information, namely for family risk assessment: to make sense of the origins of the disease in the family. | These results may help genetics healthcare practitioners understand how families perceive, respond to, and accommodate cancer risk counselling. The family context and history are relevant to participation in genetic counselling, testing, and surveillance methods. To support families in the way they share information about genetic risk among relatives. |
| Ashida 2015; USA | Cancer, and heart disease | Disseminators of family health history are often parents, grandparents, uncles, aunts, and nonbiological family members who tend to pass on information towards children, grandchildren, nieces, nephews, other nonbiological family members. | Enhancing social relationships within families may facilitate family health history communication among older adults. Encouraging older adults to act as role models has the potential to trigger a positive cascading effect within families. Efforts to facilitate dissemination within families should involve informing both older adults and their family members about the importance of mutual social exchanges. |
| Abad; 2017; Philipines | Congenital adrenal hyperplasia (CAH) | Mother (or grandmother when parents are not living together): primary disseminators of information in the family. Grandmother: share information with immediate and extended family, and non-relatives (friends and neighbours). Conduit between immediate and extended family and more distant relatives. Father: instrumental in sharing the diagnosis, limited to his side of the family (few relatives, usually siblings). | Genetic counsellors should support mothers who assume the role of being primary communicators and gatekeepers in the family. Genetic counsellors should involve and talk to the grandmothers to determine beliefs that they hold which can influence the process of communication and in framing the content of the information. |

**Table 2** (continued)

| 1st author; year; country | Inherited genetic disease | Health-related roles of the older generation | Implications on family health-management |
|---|---|---|---|
| Oliveira, 2017a; Portugal | Transthyretin-related familial amyloid polyneuropathy (TTR-PAF) | Modelling: normalize" vs. "dramatize the disease experience; "living transmitters of the disease experience"; (Not) Encouraging to carry out the pre-symptomatic test (PST): Encouraging, Supporting any decision. Providing practical support; Discouraging. (Not) Informing of the (risk of) disease, Motivating the search for disease-related information, Silencing. Supporting: Emotional support (carrier PST result); Advising on personal life decisions; Emotional and instrumental support during the disease; Supporting the decision of undergoing the available treatments. | These roles suggest the influence of older family members in the various stages of the family adaptation to the hereditary disease. Explore the interaction between older and younger family members in the context of the evolving possibilities of genomic medicine and its influence on genetic counselling. |
| Oliveira, 2017b; Portugal | Transthyretin-related familial amyloid polyneuropathy (TTR-PAF) | Mostly between women, from mother to daughter, and from older affected individuals to young pre-symptomatic carriers. Less often older males (fathers), uncles. Also, aunts and grandparents. | The involvement of older family members in family management of genetic risk can contribute to the effectiveness of the dissemination of preventive surveillance measures and healthier behaviours in family members. |
| Pantaleao, 2019, USA | Li-Fraumeni Syndrome (LFS) | Family health leaders: obtaining and disseminating new health-related information, facilitating healthcare appointments, and serving as another members' health LFS expert/advocate. Tangible or emotional support. Parents, grandparents, aunts, and uncles. Often adopted by the first identified TP-53-positive individual in the family. Most frequently a female parent with LFS if she had an affected child under 22 years. | Providers may introduce psychoeducational interventions, noting the shift in healthcare duties to younger generations and framing it as a common developmental process in families with hereditary cancer risk. Providers can tailor their approach to patient management toward family-oriented strategies of information. |
| Oliveira, 2021. Portugal | Huntington Disease (HD) | Shaping awareness about HD (promoting or hampering through providing information, awareness trigger, living transmitters of the disease, retrieved testimony, silence, denial). Influencing HD management (encouraging/supporting any decision about PST, discouraging PST, provide and receive support (emotional and instrumental), not providing support, modelling health related behaviours by normalizing or disrupting the disease experience, advocacy. | Older relatives are determinant to understand the family history and support younger generations adjustment to the disease. Genetic counselling and interventions aimed at supporting families should considerer a narrative approach specifically involving older relatives, since they have a great influence in sustaining family stories. Acknowledgement of how older generations cope with illness demands is important to understand the family's style of adjustment. |

discouragers of health screening or genetic testing; (non-) supporters; and role models in living and coping with the disease.

## Results

### Overview of the included studies

The 11 studies included (Tables 1 and 2) were published from 2009 to 2021, in the USA (5), Portugal (3), the UK (1) and Philippines (1). Three explored health-roles performed by different generations within the family (Ashida et al. 2010; Koehly et al. 2009; Pantaleao et al. 2019); five focused specifically on older family members' roles (Ashida et al. 2011; Ashida and Schafer 2015; Oliveira et al. 2017a, b, 2021); three publications did not focus on health related roles, but included relevant information on how older family contributed to the family regarding this topic (Abad et al. 2017; Lehmann et al. 2011; Mendes and Sousa 2012). Out of the 11 publications included seven were qualitative (Abad et al. 2017; Lehmann et al. 2011; Mendes and Sousa 2012; Oliveira et al. 2017a, b, 2021; Pantaleao et al. 2019) and four quantitative (Ashida et al. 2010, 2011; Ashida and Schafer 2015; Koehly et al. 2009). All quantitative studies were observational and cross-sectional. Three studies had families as participants (Abad et al. 2017; Mendes and Sousa 2012; Pantaleao et al. 2019), the other studies considered family members individually. Sample sizes in quantitative studies ranged from 110 (Ashida and Schafer 2015) to 452 (Ashida et al. 2010) participants while qualitative studies ranged from 10 (Oliveira et al. 2021) to 62 participants (Pantaleao et al. 2019). Two studies only included members from the older generations as participants (Ashida and Schafer 2015; Lehmann et al. 2011), while the nine studies included participants from the older generations. Women were predominant in the samples of all studies (three studies only included women in their samples) (Abad et al. 2017;

Koehly et al. 2009; Lehmann et al. 2011). One study focused specifically on grandmothers (Lehmann et al. 2011).

In terms of the IGC, four publications focused on hereditary cancers (Ashida et al. 2011; Koehly et al. 2009; Mendes and Sousa 2012; Pantaleao et al. 2019), one on cardiovascular disease (Ashida et al. 2010), one on cancer and heart disease (Ashida and Schafer 2015), one study focused on X-linked conditions (Fragile X and Duchenne muscular dystrophy) (Lehmann et al. 2011), two papers focused on Transthyretin-related Familial amyloid polyneuropathy (TTR-FAP) (Oliveira et al. 2017a, b), one on Huntington disease (HD) (Oliveira et al. 2021), and one on Congenital Adrenal Hyperplasia (CAH) (Abad et al. 2017).

## Characteristics of the members of the older generations

Most publications did not provide data on the characteristics of the members of the older generations. However, some studies reported older family members as being typically women, first-degree relatives, commonly parents (Ashida et al. 2010; Ashida and Schafer 2015; Koehly et al. 2009; Oliveira et al. 2017b; Pantaleao et al. 2019) and most often the mother (Abad et al. 2017; Ashida et al. 2010; Oliveira et al. 2017b; Pantaleao et al. 2019). Grandmothers were mentioned in six papers (Abad et al. 2017; Lehman et al., 2011; Ashida et al. 2010; Ashida and Schafer 2015; Oliveira et al. 2017b; Pantaleao et al. 2019). Grandparents in general, aunts and uncles were referred in four papers (Ashida et al. 2010; Ashida and Schafer 2015; Oliveira et al. 2017b; Pantaleao et al. 2019).

## Older generations health-related roles

Four main roles were identified: informers vs. blockers of disease-related information; encouragers vs. discouragers of health screening or genetic testing; (non-)supporters; and role models in living and coping with the disease.

### *Informers vs. blockers* of disease-related information

Ten publications indicated that older generations play information health-related roles toward other family members family (Abad et al. 2017; Ashida et al. 2011; Ashida and Schafer 2015; Koehly et al. 2009; Lehmann et al. 2011; Mendes and Sousa 2012; Oliveira et al. 2017a, b, 2021; Pantaleao et al. 2019). Members of the older generations gathered information about the family health history, as they often know or witnessed the circumstances of previous generations members (Koehly et al. 2009; Lehmann et al. 2011; Mendes and Sousa 2012). They also play roles as searchers for new health and risk management information from professional or support group sources (Lehmann et al. 2011; Pantaleao et al. 2019). Their roles include being disseminators of this health information towards their family members (Abad et al. 2017; Ashida et al. 2011; Ashida and Schafer 2015; Mendes and Sousa 2012; Oliveira et al. 2017a, b, 2021; Pantaleao et al. 2019). Although less often and with minor expression, four studies described older generations acting as blockers of the exchange of health information within the family. This includes being reluctant about gathering family health information or disseminating health information to family members, and being silent about the disease (Koehly et al. 2009; Oliveira et al. 2017a, b, 2021) or denying the existence of the disease (Oliveira et al. 2021).

### Encouragers vs. discouragers of health screening or genetic testing

Encouraging health screening or genetic testing is another role identified in six publications (Ashida et al. 2010, 2011; Mendes and Sousa 2012; Oliveira et al. 2017a, b, 2021). Encourager comprises assuming being favourable, booking appointments, accompanying to consultations, and advising health screening or genetic testing. In one study, this encouragement was associated with higher levels of intention to screen in other family members (Ashida et al. 2010). In three studies with rare incurable autosomal (late onset) dominant diseases, not encouraging or not supporting testing was assumed by some members of the older generations, because treatment was not available (Oliveira et al. 2017a, b, 2021).

### (Non)supporters on practical and emotional dimensions

Providing support was identified in five publications (Ashida et al. 2011; Oliveira et al. 2017a, b, 2021; Pantaleao et al. 2019), being described in various ways: a practical dimension (Ashida et al. 2011; Oliveira et al. 2017a, b, 2021; Pantaleao et al. 2019), including facilitating healthcare appointments, doing daily tasks such as preparing meals or helping in the care of children; and an emotional dimension (Ashida et al. 2011; Oliveira et al. 2017a, b, 2021; Pantaleao et al. 2019), which encompasses providing hope to other family members regarding the effect of treatments, advising on personal life decisions, or making them feel accepted. Not providing supporting was rare; however, one study described situations in which older family members have been unsupportive towards relatives affected by Huntington disease (e.g. abandoning them) (Oliveira et al. 2021).

### Role models in living and coping with the disease

Modelling behaviours was described in three studies (Oliveira et al. 2017a, b, 2021). Modelling comprised events in which the member of the older generation (usually affected by an IGC) influence the younger ones through their own positive example in how they are managing the disease. It can occur by normalizing the illness experience through e.g., trying to remain optimistic, adapting and keeping a normal life despite the disease; or through their difficulties in coping with the disease such as isolation, withdrawal, despair, and desire to die. One study suggested that modelling also occurs by being an advocate for the rare disease community, namely through involvement in patients' associations, sharing their experience with students and with the community (Oliveira et al. 2021).

## Discussion

This scoping review aimed to synthesise the health-roles performed by members of the older generations in families living with IGCs. Overall, the literature is scarce and limited to a narrow set of diseases. Hereditary cancers were the most represented IGC in the included studies. This suggests a paucity of research focusing other conditions that have traditionally received less attention than hereditary cancers.

In terms of the characteristics of the older generations, included studies show that roles are mainly performed by first-degree relatives, especially women/mothers, although fathers, grandparents, uncles, and aunts have also been identified. This is in line with literature describing mothers as the most influential members in the family network, and the role of women as gatekeepers in the management of health-related issues, including genetic risk (D'Agincourt-Canning 2001). The involvement of aunts and uncles suggests the importance of considering older individuals in the context of extended family networks beyond lineage, i.e. including biological and nonbiological members (Ashida et al. 2011; Koehly et al. 2009; Vicente and Sousa 2010).

This scoping review identified four roles: informers vs. blockers of disease-related information; encouragers vs. discouragers of health screening or genetic testing; (non) supporters on practical and emotional dimensions; and role models in living and coping with the disease. Typically, the roles performed by the older generations tend to be encouraging and adaptive. However, the included studies also report the inverse, i.e., members of the older generations who act as blockers of information, discourage genetic testing and screening, do not provide support and act as models of facing the disease with helplessness. When older generations actively engage in informing, encouraging, supporting, and normalizing role models, they become supportive allies in helping family members cope with their conditions. Nevertheless, roles involving withholding information, discouraging, and being unsupportive or a negative role model should also be carefully considered, taking into account their impact not only on the older relative but also on the entire family (Ashida et al. 2010, 2011; Ashida and Schafer 2015; Koehly et al. 2009). These varied experiences within the family may be beneficial to younger relatives in that they may gain early exposure to various coping mechanisms for dealing with the condition, enabling them to either align with or doing differently from the approaches adopted by older relatives (Oliveira et al. 2022). Also, when working with individuals and families, it is relevant to consider the broad range of interpersonal roles within the family and their implications.

The identified roles are commonly performed by older relatives and are part of family life, including caregiving for grandchildren and the provision of emotional, practical, and financial support to various relatives (Attar-Schwartz and Buchanan 2018; Barnwell 2018; Silverstein and Zhang 2020). These exchange processes involve interactions between different family members that influence attitudes and behaviour in another generation. They assume particular relevance in families facing IGCs as they often need to process complex information and involvement across and within generations (Mendes et al. 2018). For example, older relatives' privileged access to information from previous generations may be crucial to facilitate gathering and disseminating information about the family health history. In sum, our review suggests that older generations play instrumental roles that influence the younger and next generations' adaptation to the IGC. Results reinforce the relevance of acknowledging the roles of older generations from an intergenerational perspective and how they may positively or negatively influence the health-related attitudes and behaviors of younger generations in families with IGCs (Ernsth-Bravell et al. 2016; Koehly 2017).

### Limitations

Some limitations need to be considered within this scoping review. The quality of the studies has not been assessed, given the paucity of data in this topic. The authors were interested in mapping the existent research about the health-roles roles performed by the older generations in families living with inherited genetic conditions. The included studies used different settings, methods, and populations, not allowing for direct comparisons within the data. Our review is also limited by the inclusion of studies only in English and Portuguese languages and published in peer-reviewed journals. Unpublished data or data published by other means

or in other languages that could have contributed to a better understanding of the research questions were not included.

### Implications for practice and future research perspectives

Main findings in this scoping review highlight the centrality that communal and relational modes of coping have in families with IGCs (Oliveira et al. 2023). All the reviewed studies highlight the importance of acknowledging and understanding the roles of older generations for healthcare professionals, as it would contribute to a greater understanding of the family dynamics and adjustment to an IGC. This suggests these findings may be relevant to enhancing family-centered care in health care services to foster opportunities for reciprocal intergenerational interactions. Multifamily interventions are well-suited when working with families with IGCs as they maximize family engagement and promote opportunities for families to expand their social networks. These interventions have been applied in the context of chronic illness (Rocha et al. 2013) and IGCs (Mendes et al. 2010; Guerra et al. 2023), showing promising results.

Future studies addressing the roles of older generations in families living with IGCs should cover a broader range of IGCs with different features, to examine if and how roles of the older generation may differ. More clarity on these roles may shed light toward identifying which family members are better positioned to provide specific types of support to other family members according to the type of IGC. In addition, there is a lack of studies that examine the effects of the health-related roles performed by the members of the older generations on the functioning of the family and on individual members.

## Conclusions

To our knowledge, this review is the first to synthesise the information available on the health-related roles of older generations in families with IGCs. The results suggest older adults play relevant roles related to the management of the IGC in families: informers vs. blockers of disease-related information; encouragers vs. discouragers of health screening or genetic testing; (non-)supporters (practical and emotional); and role models in living and coping with the disease. Engaging members of the older generations in the health management of family's living with IGCs should be considered in order to foster reciprocal interactions and support to families and individuals.

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**Data availability** No datasets were generated or analysed during the current study.

### Declarations

**Competing interests** The authors declare no competing interests.

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