International Journal of Cardiology Congenital Heart Disease 19 (2025) 100567

Contents lists available at ScienceDirect

International Journal of Cardiology Congenital Heart Disease

journal homepage: www.journals.elsevier.com/international-journal-of-cardiology-congenital-heart-disease

Family caregiver frailty in adult congenital heart disease is overlooked: A call to action

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ARTICLE INFO

Keywords:
Congenital heart disease
Family caregivers
Frailty
Transition to adult care
Family-centered care
Health outcomes

ABSTRACT

The frailty of family caregivers (FCs) of adults with congenital heart disease (CHD) is an underrecognized yet critical issue in healthcare. As individuals with CHD live longer, their FCs, often aging alongside them, face escalating physical, emotional, and psychological challenges. This brief report examines frailty in FCs as a multidimensional issue stemming from prolonged caregiving demands and cumulative stress. The evidence gathered in this regard identifies three key domains affecting FCs: psychological stress and childcare, physical health decline, and the challenges of caregiving during transitions to adulthood. These findings underscore the urgent need for family-centered care models that incorporate tailored frailty assessments and targeted interventions to support FCs throughout their caregiving journey. Addressing caregiver frailty could improve outcomes for both patients and caregivers, strengthen resilience, and optimize resource allocation. This perspective calls for immediate action in research and policy to prioritize caregiver well-being in transition care programs, enhancing the quality of life for both caregivers and patients.

1. Introduction

The aging congenital heart disease (CHD) population and increased life expectancy present growing challenges for both individuals and their family caregivers (FCs) [1]. This is particularly relevant, as associated morbidities impair physical function, worsen health status, and drive up healthcare costs through increased service utilization [2]. These challenges extend to middle-aged and older family caregivers (FCs) who support individuals with CHD, often leading to frailty, functional decline, and adverse health outcomes in caregivers themselves [3]. This brief report brings an evidence-based perspective to elicit reflections on the grey areas still surrounding the concept and measurement of frailty in these caregivers, underscoring the need for greater clarity to support both caregiver and patient well-being in ACHD transition care programs [4].

2. The role of family caregivers in adult frailty care

Assessing frailty is essential for providing appropriate and tailored healthcare services to individuals and their families, adopting a life course approach. Thus, frailty is a predictor of healthcare expenditures throughout the entire life cycle [5]. FCs of individuals with CHD are at a particularly high risk of developing frailty in middle and older age [1,3]. As reported in a recent study by Kiljunen, approximately 70 % of informal family caregivers were identified as frail [6]. Additional studies have shown that family functioning and the well-being of FCs are significant predictors of frailty in older adults with chronic illness [7]. Given this context, shifting from a patient-centered to a family-centered

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https://doi.org/10.1016/j.ijcchd.2025.100567
Received 8 January 2025; Received in revised form 12 January 2025; Accepted 13 January 2025
Available online 17 January 2025

assessment is fundamental for effectively managing and preventing frailty as a "family affair", especially within the population with CHD. In this regard, a recent scientific statement by the American Heart Association highlights the pivotal role and engagement of family members in adult cardiovascular care, aiming to achieve better outcomes for both patients and their FCs [3]. Considering these findings, similar reflections and initiatives are also needed within the European context to comprehensively address the needs of FCs and promote optimal care for aging populations [4].

3. Frailty in the population with congenital heart disease

The definition of frailty has reached a common international consensus: it is characterized by a functional decline in physical, psychological, or cognitive systems coupled with increased vulnerability to stressors [8]. Although frailty is closely correlated with aging, it can occur at any age [5,8]. Therefore, the presence of biomedical risk factors in midlife is often linked to frailty in later years, a phase known as "prefrailty." This multifactorial, multidimensional state involves physiological and adverse processes that evolve over time. Consequently, both frailty and prefrailty are associated with an increased risk of disability, morbidity, falls, hospitalization, and mortality [5,8].

In individuals with CHD, the prevalence of frailty and prefrailty has been studied in both adult and pediatric populations. Recent results from the APPROACH-IS II trial revealed that among middle-aged and older adults with CHD, 41.9 % were classified as prefrail, with 5 % assessed as frail. Furthermore, more than one-third experienced cognitive impairment. Factors such as clinical conditions, age, sex, physiological class, and socioeconomic status were identified as predictors of frailty in this group [1].

Some limited evidence also suggests that children and adolescents with CHD or acquired heart disease exhibit lower physical capabilities and poorer psychological well-being compared to their healthy peers [9]. However, to the best of our knowledge, the frailty or prefrailty phenotypes among FCs of individuals with CHD have been largely overlooked. This gap currently limits our understanding of how FCs cope with and experience the demands of supporting individuals with CHD as they transition through different life stages—from emerging adulthood to middle and late adulthood.

4. What are the key literature topics on family caregivers in the population with CHD?

In October 2024, we gathered evidence from the literature to identify the main topics addressed in the literature on FCs of individuals with CHD. We consulted the PubMed database and secondary sources from grey literature (see Appendix 1 for the search strategy). Articles published in English or Italian within the last ten years focusing on the population with CHD and addressing family caregiving practices were considered eligible for gathering the required literature. There were no restrictions on study design. However, we excluded studies focusing on topics such as end-of-life care in paediatrics, caregiving for newborns, and life-threatening cases to guarantee consistency with the idea of gathering diverse sources of evidence from the literature to identify the main topics regarding FCs of individuals with CHD. Two authors conducted the study selection by screening titles and abstracts, identifying 54 studies for the eligibility phase. After reviewing the full texts of the eligible studies, 17 were included in the textual analysis. Data extraction focused on the research purpose, sample, main results, and conclusions for each included study. All extracted information is summarized in Appendix 2.

For the textual analysis of the gathered literature, we used Latent Dirichlet Allocation (LDA), a topic modeling approach, to identify latent topics through a textual lexicometry method. Lexicometric characteristics of the analyzed textual corpus are shown in Appendix 3 and suggested suitability of topic modeling. Additionally, multiple correspondence analysis (MCA) was performed to examine how these topics were distributed by variables such as country and years of publication (see Appendix 3).

As shown in Fig. 1, three main topics emerged (fit indices as shown in Appendix 3): (1) psychological support and child care, (2) parental stress and physical health, and (3) adult care and emotional response. The first two topics, "psychological support and child care" and "parental stress and physical health," make up 82.4 % of the topic distribution, indicating a primary focus on childhood care. Notably, caregiving for newborns, infants, and adolescents with CHD significantly impacts the physical and mental health of FCs, affects the family system as a whole, and also influences the child's development. Research estimates that up to 30 % of parents of children with CHD exhibit symptoms of post-traumatic stress disorder, 25–50 % show symptoms of anxiety and depression, and 25–50 % experience psychological distress. Sleep disorders, lack of energy, fatigue, and lack of energy are common, impairing FCs' ability to manage household duties and social activities. Older family caregivers, in particular, often neglect their own health needs due to time constraints). Financial strain and social restrictions further diminish FCs' quality of life).

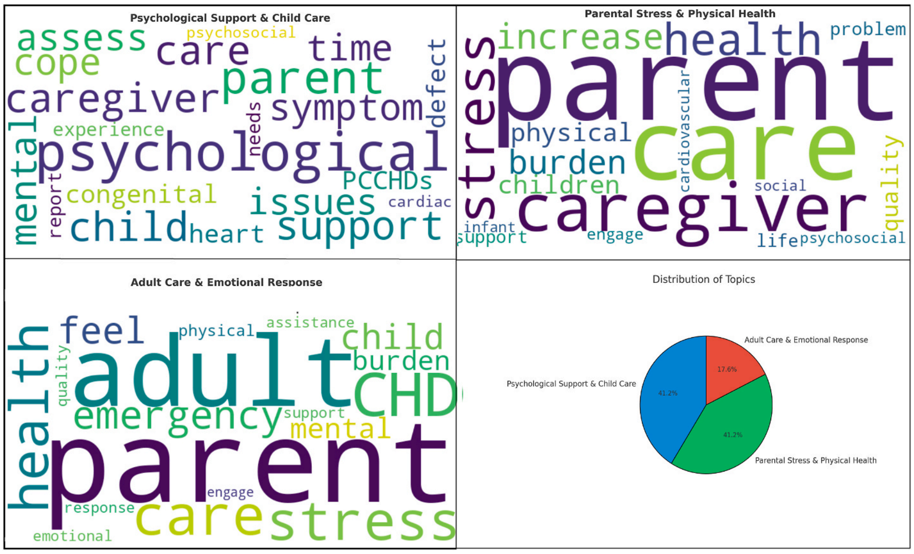
The third topic, "adult care and emotional response," represents only 17.6 % of the distribution. Limited evidence suggests that caring for adults with CHD (ACHDs) remains a substantial challenge for FCs, who often take on an advocacy role. FCs experience uncertainty and emotional stress related to diagnosis, family planning, employment opportunities, and ACHDs' ability to navigate the healthcare system and independently manage their health, particularly for those with cognitive or physical impairments. Furthermore, financial burdens and difficulties in maintaining health insurance can lead to prolonged dependence on family support for ACHDs.

Even though the evidence gathered to perform the LDA is not as comprehensive as it could be in a full systematic review, it provides valuable insight into the current research landscape on family caregivers of individuals with CHD across different life stages. The gathered evidence in this brief report helps to clarify the issues at hand and paves the way for more in-depth, stand-alone reviews on this important subject by highlighting key topics and challenges.

5. Understanding frailty in family caregivers of adults with CHD: Implications for long-term care

The limited evidence on FCs of ACHDs hinders the development of comprehensive, consistent, and personalized family-centered care during the transition into adulthood [3]. Viewing frailty through the lens of the cumulative deficit model by Rockwood (2016) highlights frailty as a result of accumulated health problems and life-long risk factors [5,8]. From this perspective, we propose that FCs of individuals with CHD may themselves be considered frail. Given the mental, physical, and social stressors faced in caring for a child with CHD, the reserve capacity of FCs is likely depleted by the time the individual reaches adulthood [10].

As a practical example, if the current mean age of adults with CHD is around 52 years, and the mean age of parents of children with CHD is 35 years, the mean age of FCs caring for CHD adults could now be over 80 years. Recognizing frailty and prefrailty in these older caregivers is essential for delivering sustainable health, psychological, and social interventions that mitigate adverse outcomes and related costs. This trend will become increasingly significant as life expectancy for individuals with CHD rises, alongside the broader demographic shift towards an aging population—especially within Europe. In other words, we are likely to see a higher prevalence of frailty or prefrailty among older family caregivers of ACHDs, although this data is currently underreported.



Psychological Support & Child Care

Parental Stress & Physical Health

Adult Care & Emotional Response

Distribution of Topics

Fig. 1. Identified Topics and keywords. Word clouds for three identified topics: Psychological Support & Child Care (top-left), Parental Stress & Physical Health (top-right), and Adult Care & Emotional Response (bottom-left). Keywords represent the most frequent and relevant terms associated with each topic. The pie chart (bottom-right) illustrates the distribution of topics

6. Advancing research and support for family caregivers of adults with congenital heart disease

Several key areas require further exploration to address the complex and evolving needs of FCs of ACHDs. Firstly, it is crucial to investigate how frailty develops and progresses in FCs of individuals with CHD as they age, particularly in comparison to caregivers of those with other chronic conditions. Understanding the factors that predict these caregivers' transition from prefrailty to frailty could shed light on the specific stressors and protective elements that influence caregiver resilience and vulnerability. Additionally, the impact of family dynamics and broader social support networks on caregiver well-being remains underexplored and warrants more attention.

A significant challenge in advancing this field is the identification of effective tools for measuring frailty among family caregivers. Existing frailty scales are generally designed for older adults or clinical populations and may not fully capture the unique demands associated with caregiving for individuals with CHD [1]. Determining whether these tools should be adapted specifically for caregivers or if new measurement approaches should be developed is essential for accurately assessing frailty in this context.

Another area of interest is the evolving role of significant others in the caregiving network of ACHDs. As individuals with CHD age, spouses, siblings, or close friends may increasingly assume caregiving responsibilities, raising important questions about family dynamics [3]: ≪At what point, and under what conditions, does the parental caregiver remain the primary support figure, especially as they themselves grow older? Moreover, what is the readiness of ACHDs in providing care for older family caregivers?≫ In this scenario, exploring how family-centered care could be implemented from an early stage may help prepare family members for these transitions, ensuring that ACHDs have a supportive network that adapts to their evolving needs. This approach could foster resilience and continuity in caregiving, reducing the strain on aging parents and promoting better long-term outcomes for individuals with CHD.

Encouraging family-centered care from the outset could have substantial benefits for both caregivers and the CHD population [4]. Early integration of family support into the caregiving framework may lead to healthier family dynamics, facilitate the adult development and autonomy of individuals with CHD, and enable a smoother transition in caregiving roles over time. Addressing the quality of life of all caregivers in this network—including parents, siblings, and partners—will be essential for developing sustainable caregiving practices and improving outcomes for ACHDs. Ultimately, these considerations underscore the importance of adopting an integrated, family-centered approach to care and suggest promising directions for future research and intervention.

7. Conclusion

Addressing FC frailty through tailored assessments and interventions is critical for sustaining caregiving practices, enhancing family functioning, and improving outcomes for individuals with CHD. This requires urgent integration into policy and care frameworks Future research should address existing gaps by exploring the development of frailty in FCs, defining the phenotype of frailty, creating effective assessment tools, and designing interventions to support caregivers throughout their journey. Given the global trend of an aging population, addressing the needs of family systems with older caregivers is essential for developing and investing in long-term strategies, such as transition care programs in adulthood, to foster resilience within caregiver networks and enhance the quality of life for both family caregivers and those in their care.

CRediT authorship contribution statement

Arianna Magon: Writing – review & editing, Project administration, Data curation, Conceptualization. **Serena Francesca Flocco:** Writing – original draft, Supervision, Methodology, Investigation, Formal analysis, Conceptualization. **Laura Schianchi:** Writing – review & editing, Supervision, Methodology. **Massimo Chessa:** Writing – review & editing, Supervision, Methodology. **Daniele Ciofi:** Writing – review & editing, Supervision, Methodology. **Salvatore Angileri:** Writing – review & editing, Supervision, Methodology. **Maddalena De Maria:** Writing – review & editing, Supervision, Methodology. **Gianluca Conte:** Writing – review & editing, Supervision, Methodology. **Silvia Favilli:** Writing – review & editing, Supervision, Methodology. **Cristina Arrigoni:** Writing – review & editing, Validation, Supervision, Methodology. **Rosario Caruso:** Writing – review & editing, Supervision, Project administration, Conceptualization.

Non-conflict statement

The authors declare no conflict of interest related to this work, other than MC serving the IJCCHD Editorial Board, albeit not involved in the handling of the paper.

Funding

This study was partially supported by Ricerca Corrente funding from the Italian Ministry of Health to IRCCS Policlinico San Donato.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijcchd.2025.100567.

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