



Comprehensive care for haemophilia: A literature review for improving institutional cooperation

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ABSTRACT

This paper investigates how the notion of comprehensive care (CC) in haemophilia is presented in institutional guidelines and specialized understandings. A literature review of the articles published in Haemophilia was carried out. The main features which authors refer to when mentioning CC were outlined and compared with the principles, assumptions and recommendations set by the World Federation of Haemophilia in relation to this care model. The results show a fragmented scenario. First, very few articles are devoted/explicitly refer to CC. Second, only a few of them completely adhere to the notion of CC as proposed in institutional guidelines. In contrast, most articles stress a single feature, providing a partial view of the model. The focus is mainly on promoting physical health, which is however frequently associated with psychosocial health. Multidisciplinarity has great emphasis, too. Third, some key issues for CC emerge. They are implicitly present in institutional guidelines, but they become the very focus of some articles, which reflect on three challenges to be dealt with: care for all, transition care and systemic care. Such challenges offer the opportunity to clarify and expand the notion of haemophilia CC, and to propose future developments in research projects and interventions.

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Introduction

State of the art

To date, knowledge on haemophilia, as well as bleeding disorders at large, among non-specialist healthcare professionals (HCPs) is quite limited since it is a rare disease. As a consequence, both haemophilia diagnosis and management in emergency situations are complex when conducted outside specific hospital departments.

In recent years, the greater number of therapeutic options and medical devices developed by the pharmaceutical industry and relying on biomedical research and multidisciplinary theoretical approaches have reduced mortality and increased the quality of life of people with haemophilia (PWH). There are, however, considerable disparities between countries due to social, economic, political and cultural differences that affect the amount, availability and type of treatments proposed, as well as the use of a more or less integrated approach to the definition of health and care.

Notion of comprehensive care in institutional guidelines

Today, the care model promoted by the World Federation of Haemophilia (WFH), and set by the guidelines recently revised after their first edition in 2005 [1], is

known as 'comprehensive care' (CC) (Figure 1). One of its principles is that PWH are best managed in a CC setting. In particular, the model is based on two main assumptions. First, it stresses the significance of jointly promoting physical and psychosocial health, while improving the quality of life of PWH and their families, and decreasing morbidity and mortality. Second, CC emphasizes the necessity of co-ordinately combining different HCPs into multidisciplinary teams that are responsive to the wide-ranging needs of PWH and their families. CC teams should be able to protect the physical and psychosocial health of PWH and their families, thus they should include medical directors, nurse coordinators, musculoskeletal experts, laboratory specialists, psychosocial experts, and should be the reference point in order to approach different non-specialized HCPs (e.g. dentists, geneticists, gynaecologists).

These two assumptions translate into concrete actions as well. A CC programme should provide continuous, long-term and multidisciplinary support to PWH and their families [e.g. 2]. This involves the identification of resources and strategies to help cope with everyday risks and problems, specificities of different life stages, school and/or employment issues, and reproduction options, in accordance with specific national treatment guidelines. In other words, this implies ensuring prompt management of bleeding

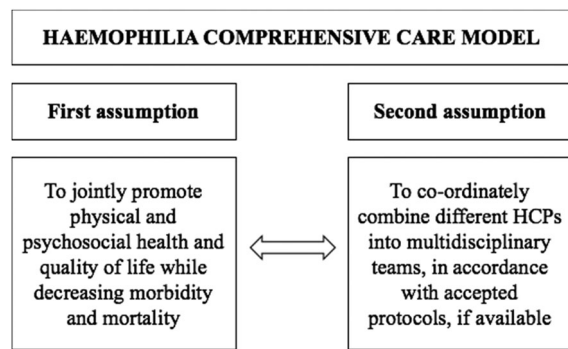


Figure 1. Diagram of the comprehensive care model (adapted from [1]).

events, prevention of (potential) related complications and attention to psychosocial health at the haemophilia comprehensive care centres (HCCCs) [e.g. 3,4]. Moreover, a CC programme should guarantee full training and close supervision of home treatments among PWH, as well as adequate tutoring for their families to meet the needs of PWH in everyday life [e.g. 5,6].

Rationale of the paper

The aim of this paper is to examine the mutual relationship between the notions of haemophilia CC proposed by institutional guidelines and specialized understandings. More precisely, this paper intends to investigate the implementation of the institutional guidelines in everyday medical practices and, vice versa, the capacity of HCPs to set the institutional agenda. For this purpose, starting from a systematic review of the articles published in the *Haemophilia* journal, it will illustrate the main issues proposed by the academic literature in dealing with this care model. These issues will be framed according to the institutional guidelines set by the WFH, in order to identify the overlapping between scientific sources and WFH guidelines. This literature review will lead to an extensive theoretical reflection on this topical issue and will allow us to identify and emphasize neglected and/or emerging aspects that might be relevant for future research and interventions.

Method

The review of currently available scientific literature was conducted by applying the following three-step procedure (i.e. search, qualitative synthesis, discussion), inspired by the Search, Appraisal, Synthesis and Analysis (SALSA) analytic framework.

The first step regarded the search procedure. The review focused on the *Haemophilia* journal, since this international periodical publication is the official journal of the WFH, thus representing an influential venue

for providing readers with an overview of the current state of the art on haemophilia and bleeding disorders at large. Moreover, the aim of this review is in line with the Journal's scope: *Haemophilia* is 'dedicated to the exchange of information regarding the comprehensive care of haemophilia'.¹ The main goal of the current paper was to explore how the articles published in *Haemophilia* apply and describe the notion of CC: that is, whether this notion is coherent with the institutional guidelines previously outlined or, by contrast, it proposes partial or specific views. All the 4,446 articles published since 1995, when *Haemophilia* was founded, to the end of 2016 were considered for this review. In particular, these articles were selected according to the presence of the words 'comprehensive' and 'care' within the title or abstract.

Then, these documents were assessed according to the 'relevance' of the notion of CC developed within each article. To be specific, they were categorized into 'high relevance articles' (i.e. the article is explicitly devoted to CC), 'medium relevance articles' (i.e. the article gives definitions, explanations or information about this notion) and 'low relevance articles' (i.e. the article only uses the term CC without adding any definitions, explanations, or information).

Finally, medium and high relevance articles were included in the subsequent qualitative synthesis and discussion aimed at identifying and commenting the main features of CC proposed by the authors.

Results

General information

The search procedure was completed following four steps, which are graphically summarized in Figure 2.

CC was mentioned in titles and/or abstracts in 80 articles, which constituted our initial corpus, distributed in volumes and years as illustrated in Figure 3. Figure 3 shows the irregular, but generally increasing, trend of articles that explicitly referred to CC over time.

Relevance of comprehensive care

An eligibility screening of the corpus led us to assess the articles according to the relevance of the notion of CC and to exclude 33 low relevance articles, where the notion of CC was only mentioned rather than actually developed throughout the entire paper. Thus, the retained corpus consisted of 47 articles, which were included in the subsequent qualitative analysis and discussion: 36 medium relevance articles and 11 high relevance articles. These three categories of articles were distributed discontinuously over time; however, the frequency of medium and

¹cf. [http://onlinelibrary.wiley.com/journal/10.1111/\(ISSN\)1365-2516/homepage/ProductInformation.html](http://onlinelibrary.wiley.com/journal/10.1111/(ISSN)1365-2516/homepage/ProductInformation.html)

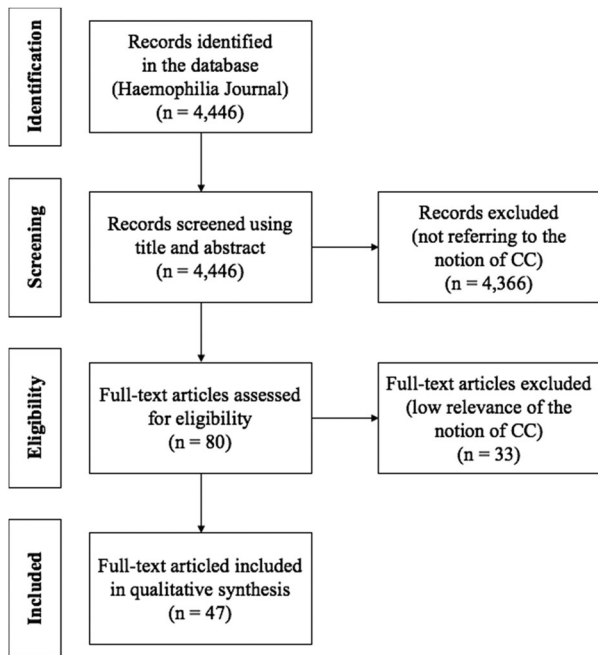


Figure 2. Review PRISMA flow diagram.

high relevance articles increased within the most recent volumes (Figure 3).

Notion of comprehensive care in specialized understandings

A qualitative analysis of medium and high relevance articles showed that only a few of them ($n = 7$) adopt the notion of CC consistently with the institutional guidelines. In contrast, the majority of articles ($n = 24$) focuses on a single feature of this notion, thus providing a partial view of the model. Lastly, some papers

($n = 16$) discuss specific issues related to the construct of CC. Table 1 summarizes the main features of CC considered in the analyzed articles, providing a comparison between what is proposed in institutional guidelines and in specialized understandings.

Complete correspondence with the definition of CC in institutional guidelines

The articles adhering to a complete definition of CC can be classified into two groups.

Overviews. This group consists of four articles providing an overview of CC principles. This is the case of two seminal works by Evatt [7,8], which are specifically devoted to the notion of haemophilia CC. In particular, Evatt et al. [7] illustrate how this care model is adopted in – and adapted to – different geographical contexts around the world; Evatt [8] discusses the challenges to be faced in order to fully develop and sustain CC globally. In a similar vein, Skinner [9] stresses the importance of ensuring continued global collaboration and advances on the research front to reduce the care gap between developed and developing countries. Lastly, Colvin et al. [10] set out the principles of haemophilia CC by focusing on the European context.

Case studies. This group consists of three articles that apply the definition of CC to specific case studies. Teixeira et al. [11] show how national and local haemophilia registries could represent valuable tools to ensure CC, as they support HCPs, empower PWH and improve communication. Escobar et al. [12] describe the best practices developed and adopted by HCCCs to adhere and extend the CC model. More recently, Zia et al. [13] highlight the need to offer CC to women with heavy menstrual bleeding and blood

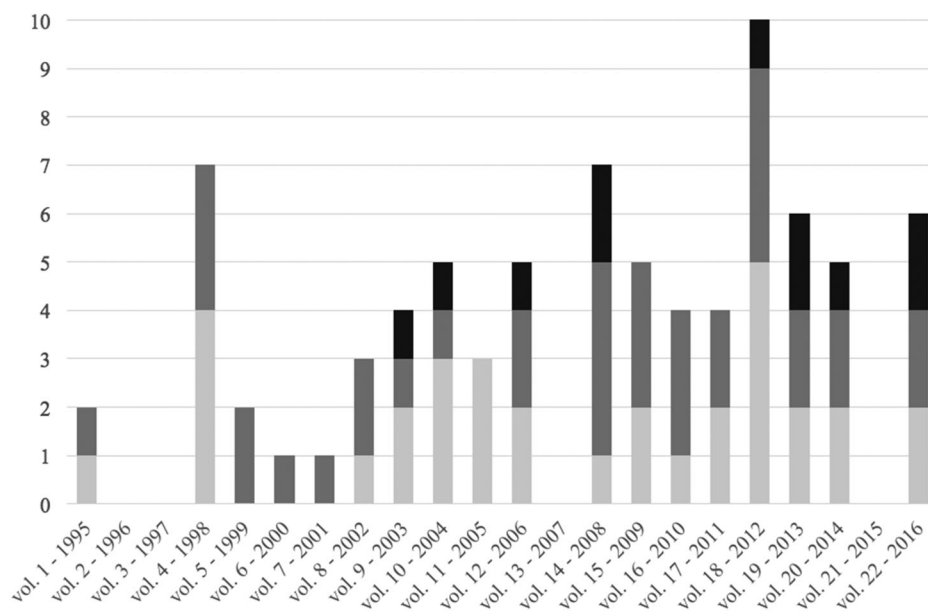


Figure 3. Trend of articles that mention the notion of 'comprehensive care' by volume/year and relevance.

Note: The X axis indicates volumes and years; the Y axis indicates the number of articles in which the notion of comprehensive care has low (light grey), medium (dark grey) and high (black) relevance.

disorders as well as to design specific programmes in this direction.

Partial correspondence with the definition of CC in institutional guidelines

The articles providing a partial view of CC could be classified into several groups as well.

Focus on the first assumption of CC. This group includes six scientific contributions especially interested in the first assumption underlying the notion of CC, that is, the importance of jointly promoting physical and psychosocial health. In this group, the gender viewpoint again comes to the forefront. Kouides [14] and Yang and Ragni [15] discuss how testing and care provision in HCCCs may empower women with bleeding disorders to seek medical care and improve their quality of care. Tedgård [16] and Miller [17] underline how genetic counselling, carrier testing and prenatal diagnosis of haemophilia, as well as their psychological consequences, represent a main dimension of CC, since they could enable conscious personal decisions concerning marriage and childbearing. Furthermore, Elander and Barry [18] address the topic of pain and its management in haemophilia. Lastly, Wodrich et al. [19, p. 593], who state that ‘comprehensive care of boys with haemophilia must include awareness of his psychosocial and physical health status’, investigate the hypothesis that boys with extreme levels of attention deficit hyperactivity disorder (ADHD) symptoms are over-represented among boys with haemophilia.

Focus on the first assumption of CC, especially on physical health. This group is composed of five articles that underlined the first assumption as well, but with a particular interest in physical aspects rather than in psychosocial ones. Evatt et al. [20] and Berntorp et al. [21] focus on treatments. They stress the need to exactly define when and which prophylaxes should be adopted, as well as to periodically re-evaluate and adjust the adopted management techniques and used therapeutic products. Moreover, surgeries are the main topic of the contributions proposed by Rangarajan et al. [22] and Rodriguez-Merchan [23]. These papers discuss the fact that a wide variety of surgeries can be successfully performed in adult PWH within a CC setting and describe the standards for the correct care of post-operative wounds. Lastly, Salem and Eshghi [24] address oral and dental health. They stress the long-term beneficial effects for patients with bleeding disorders of receiving supportive oral and dental care at a young age.

Focus on the first assumption of CC, especially on psychosocial health. Only two articles precisely emphasize the promotion of psychosocial health, without taking into account the other aspects proposed in institutional guidelines. This is the case of Wiedebusch et al. [25], who discuss the importance of psychosocial care

at family level, by reducing psychosocial strains and enhancing the adaptive coping strategies. More recently, Iannone et al. [26] advocate for the incorporation of depression screening and treatment as part of CC for adult PWH.

Focus on the second assumption of CC. This group gathers the five articles focused on the second assumption underlying the notion of CC, that is, the need to co-ordinate different HCPs into multidisciplinary teams. Cahill et al. [27], who mention the notion of CC for the first time, and Kuhathong et al. [28] focus on the role of nurses within a multidisciplinary team in HCCCs. These authors discuss how nurses act as both CC providers for patients and as trainers for patients and their caregivers. Heijnen et al. [29] provide an overview of the multiple roles played by the physiatrists as members of multidisciplinary teams in different Countries, including transversal education, specialized treatment and PWH involvement. Poon and Luke [30] highlight the importance of continuative training for multidisciplinary HCPs in order to develop haemophilia CC with emphasis on service, education and research. Lastly, Mauser-Bunschoten et al. [31] address the need for unique, consistent and exhaustive documentation concerning the patients’ status and history: this document should be filled out, consulted and shared among all the members of CC teams.

Focus on both assumptions of CC, especially on physical health. Five articles stress both the above-mentioned assumptions, but focus on the need for a multidisciplinary approach for the promotion of physical health, ignoring any psychosocial purpose. De Kleijn et al. [32], Giangrande et al. [33] and Kulkarni [34] address the topic of surgeries. They argue that surgeries should only be undertaken in HCCCs, which have the required multidisciplinary experience and facilities, and illustrate a number of coordinated procedures carried out by multidisciplinary teams to ensure optimal outcomes before, during and after surgeries. Dargaud and Negrier [35, p. 228] describe the characteristics that a HCCC should possess: ‘collaboration between the widest possible range of specialist staff, 24-h access to medical expertise, laboratory technology for the diagnosis and management of haemophilia, and appropriate and sufficient treatment products’. More recently, Kulkarni et al. [36] suggest that the use of telemedicine may become a useful tool to devise, deliver and monitor multidisciplinary CC to patients.

Focus on both assumptions of CC, especially on psychosocial health. Only one article focuses on both the assumptions with specific emphasis on the promotion of psychosocial health, rather than considering issues related to patients’ physical health. Coppola et al. [37, p. 26] claim that providing CC, counselling and support to PWH and their caregivers in HCCCs has always been problematic for practical and psychological reasons. In this regard, the authors state that

Table 1. Summary of the main features characterizing the notion of comprehensive care in medium (lower cases) and high (upper cases) relevance articles.

	First assumption – Physical health	First assumption – Psychosocial health	Second assumption – Multidisciplinary team	Specific issues
EVATT ET AL. [7]	*	*	*	
EVATT [8]	*	*	*	
Skinner [9]	*	*	*	
Colvin et al. [10]	*	*	*	
Teixeira et al. [11]	*	*	*	
Escobar et al. [12]	*	*	*	
Zia et al. [13]	*	*	*	
Kouides [14]	*	*		
Yang and Ragni [15]	*	*		
Tedgård [16]	*	*		
Miller [17]	*	*		
ELANDER AND BARRY [18]	*	*		
Wodrich et al. [19]	*	*		
Evatt et al. [20]	*			
Berntorp et al. [21]	*			
Rangarajan et al. [22]	*			
Rodríguez-Merchan [23]	*			
Salem and Eshghi [24]	*			
Wiedebusch et al. [25]		*		
Iannone et al. [26]		*		
Cahill et al. [27]			*	
Kuhathong et al. [28]			*	
HEIJNEN ET AL. [29]			*	
Poon and Luke [30]			*	
Mauser-Bunschoten et al. [31]			*	
De Kleijn et al. [32]	*		*	
Giangrande et al. [33]	*		*	
KULKARNI [34]	*		*	
Dargaud and Negrier [35]	*		*	
Kulkarni et al. [36]	*		*	
Coppola et al. [37]		*	*	
Srivastava et al. [38]				*
Santiago-Borrero et al. [39]				*
Isarangura [40]				*
TANG ET AL. [41]				*
Duncan et al. [42]				*
ALEDORT [43]				*
Oldenburg et al. [44]				*
Dolan et al. [45]				*
SMITH ET AL. [46]				*
Breakey et al. [47]				*
PAGE ET AL. [48]				*
Buzzi et al. [49]				*
PRITCHARD AND PAGE [50]				*
Calizzani et al. [51]				*
Hacker et al. [52]				*
GROGAN ET AL. [53]				*
TOTAL	23	16	18	16

Note: Articles are reported following the presentation order in the text.

the joint analysis of the psychological burden of haemophilia, with the identification of needs arising from clinical problems in every stage of life, should be viewed as the common background for the group of haemophilia specialists and psychologists working together with two goals: developing a network of psychological support services in some Italian haemophilia centres, and promoting specific educational programmes for training professionals in haemophilia comprehensive care.

Emerging aspects with respect to the definition of CC in institutional guidelines

Finally, the articles proposing specific issues in CC can be gathered into three main perspectives. Starting from the notion of CC as proposed in institutional guidelines, these viewpoints could be labelled as: ‘care for all’, ‘transition care’, and ‘systemic care’. Although

these tags are somehow included in the previous groups of papers, since they implicitly represent transversal assumptions underlying this care model, they explicitly become the central core of this last set of articles.

Care for all. Six papers proposed the ‘care for all’ view. Srivastava et al. [38] address the issue of haemophilia management in developing countries. The authors provide an overview of the main problems which emerged: along with the low level of social awareness, these difficulties entail the inadequateness of the facilities and the lack of products for therapy and their costs. Non-affluent countries are the focus of two other articles as well. Santiago-Borrero et al. [39, p. 389] state that ‘the present paradigm of haemophilia care is the comprehensive care approach, which includes a well-coordinated team of health pro-

professionals providing comprehensive continuous services and support to the patients and their relatives'. Services and support are discussed in terms of diagnosis, the assessment of the needs of PWH and collaboration among HCPs. However, this paradigm requires high costs which are not usually accessible across all countries. Isarangkura [40] highlights the need for increasing the number and the quality of facilities, as well as ensuring the continuity in product availability in such facilities. More recently, these points are also underlined by Tang et al. [41], who discuss how CC should be improved in China. These scholars stress the importance of establishing many more facilities, which should be able to rely on adequate infrastructures and CC teams. Availability and affordability of the products are considered as well. Developed countries, and specifically the United States of America, are the focus of two other articles. Duncan et al. [42, p. 519] address cost-management strategies. The authors discuss how 'to curb the cost of specialty pharmaceuticals in conditions when their expenditures represent a significant portion of total annual costs of care'. Similarly, Aledort [43] concludes his historical review with the current challenges for haemophilia CC, including its maintenance despite decreased funding.

Transition care. This group fosters the 'transition care' view ($n=4$). Oldernburg et al. [44] explicitly take an age-related perspective and state that, as a consequence of the growing life expectancy of PWH, the spectrum of current CC should be extended to diseases generally considered as a prerogative of elderly patients. In the same issue, specifically devoted to transition care, Dolan et al. [45] take the same stance. The authors stress that the delivery systems of medical CC for PWH require extensive modification in order to provide appropriate services for the management of the diseases of older age. More recently, Smith et al. [46, p. e4] further underline this point, by stating that 'the comprehensive care team needs to adapt to the changing needs of the ageing adult with an inherited bleeding disorder'. Differently, Breakey et al. [47] focus on adolescence and its challenges, that result in corresponding additional issues for haemophilia CC. Since institutional guidelines do not sufficiently foster a conscientious approach to transition care, the authors suggest significantly promoting further research in this specific field.

Care for all and transition care. Two recent articles merged the 'care for all' and 'transition care' views. On the one hand, Page et al. [48, p. 535] provide an assessment of CC programme services and resources in Canada. The authors conclude that 'many programmes have serious human resource shortcomings in their core disciplines. As a result, some programmes are unable to fully respect key standards of care'. They also add that 'this situation is exacerbated by a new and

little studied problem, the ageing of people with bleeding disorders. The co-morbidities of ageing are increasing the complexity of care and treatment, with the potential to significantly increase demands on services'. On the other hand, Buzzi et al. [49, p. e320-321] state that 'in recent years, public funding for haemophilia support programmes [...] has been reduced across many countries, placing greater responsibility on the community to drive local initiatives'. Therefore, the authors emphasized that 'more community-led programmes are needed to further empower PWH and their families worldwide, with the aim of providing comprehensive support throughout all life stages'.

Systemic care. Four articles suggested the 'systemic care' view. Pritchard and Page [50], as well as Calizzani et al. [51], translate this view in terms of 'multi-level care'. Specifically, Pritchard and Page [50] point out that CC should involve a variety of actors (e.g. organizations, institutions, community agencies, governments) working together in the continuum of care; in other words, it should fully reflect the broader health-care system through a vertical integration process. Calizzani et al. [51] recommend that the principles underlying haemophilia CC should be considered for implementation within local policy planning and organizational frameworks. Differently, Hacker et al. [52], as well as Grogan et al. [53], interpret the 'systemic care' view in terms of 'bottom-up care'. In particular, Hacker et al. [52] call for broader PWH involvement to helpfully guide decisions concerning CC services. Grogan et al. [53] advocate that the role of PWH in service quality development is crucial. The authors conclude that a model of PWH collaboration within haemophilia services should be suitable for the evaluation, planning, development and delivery of care.

Discussion

The current literature review makes it possible to draw some closing remarks about similarities and differences between the notions of haemophilia CC in institutional guidelines and specialized understandings.

Concerning institutional guidelines, haemophilia CC has increasingly become a milestone among the existing care models over time. Consequently, this notion has been defined following precise principles, constantly updated, and corresponding recommendations and practices. These guidelines seem to play an essential role: they drive the actors involved (i.e. PWH, caregivers, HCPs, but also organizations and institutions) toward the optimal management and care of this rare, chronic and complex disease. The continuous search for excellence in the quality of treatments provided represents a common challenge that characterizes health systems, not only in the haemophilia field. Recent studies have defined this challenge

as increasingly impelling over time due to demographic and epidemiological transformations underway. Many of these transformations reflect some of the specificities of haemophilia: in particular, literature has been focusing on the importance of promoting a model of integrated care for rare diseases [54] and for chronic diseases, which are constantly increasing [55–57], especially those at risk of co-morbidity [58,59].

As regards specialized understandings, the emerging scenario is more fragmented.

First, it should be noted that very few articles explicitly refer to CC, and that they are rarely devoted to this recognized care model. This lack of reference may be interpreted in a twofold way: the notion of CC may be taken for granted or, on the contrary, it may represent an over-multifaceted care model, which is thus difficult to be exhaustively implemented in everyday medical practices and discussed in literature. This last interpretation is consistent with recent literature dealing with defining integrated care, highlighting the difficulty of proposing a consensual and univocal definition [60–62].

Moreover, in line with the previous point, only a few articles completely adhere to the notion of CC as proposed in the institutional guidelines. In contrast, some gaps between institutional guidelines and specialized understandings can be highlighted.

Most articles focus on a single feature of this notion, thus providing a partial view of CC. The emphasis is mainly on promoting physical health, which is however commonly associated with psychosocial health. This reflects the more recent and increasing interest of scholars in the psychosocial issues faced by PWH and their families, shown by many empirical studies aimed at highlighting subjective experiences in everyday life [e.g. 63], or assessing the impact of psychological and socio-educational interventions implemented in collaboration with HCCCs [e.g. 64–67].

Nevertheless, while the psychosocial dimension of haemophilia has been widely treated in literature with regard to PWH and their families, the psychosocial experience of professionals working in this field has been only marginally explored. By exploring the literature tackling this issue, limited production has been identified, and this mainly focuses on the risks of burn-out due to the treatment of a chronic illness potentially subject to complications and co-morbidities [e.g. 68–70]. Yet, psychological literature has indicated that an increased awareness of the psychosocial aspects of work can improve organizational well-being, encourage increasing attention to multidisciplinary teams and teamwork, and reduce the incidence of psycho-emotional difficulties in HCPs, thereby improving the quality of care [e.g. 71]. For instance, the current review indicates that those papers focusing on multidisciplinary teams highlight the potential impact of several psychosocial issues related to the CC model, such as the

need to be coordinated, to assume new roles, and to share information. These issues imply corresponding skills, which are transversal across various professional roles, but are also expressed and contextualized depending on the specific duties and responsibilities pertaining to each role. Such skills – defined as soft skills in the field of work and organizational psychology [72,73] – are thus pivotal in order to achieve a CC approach, and may be integrated in forthcoming research designs, tool development, training paths and may orient how to organize work in HCCCs.

In addition, the notion of CC that emerged from the literature review puts at the forefront some specific issues. These issues are implicitly included in institutional guidelines as well, but they become the core topic of several articles.

Therefore, starting from the institutional definition of CC, these articles explicitly reflect on three current challenges that should be carefully handled: ‘care for all’ (i.e. the accessibility to care, both in terms of suitability/reachability of facilities and affordability/availability of treatments), ‘transition care’ (i.e. attention to specific age-related issues), and ‘systemic care’ (i.e. inclusive care, both in terms of involving all the institutional actors included in the healthcare system and promoting the participation of PWH in decision-making processes).

This result is coherent with a previous study reporting the results of a lexico-metric analysis of interviews with HCPs working with haemophilia across different countries, with the main purpose of mapping their shared cognitive, affective and behavioural experiences [74,75]. This research pointed out the recurrence of relevant themes that overlap to a certain extent with the challenges identified in the present literature review. Specifically, along with communicating diagnosis and counselling, and considering the relational networks of PWH, the following topics arise: dealing with policies, resources and stakeholders in the local care system (i.e. ‘care for all’ and ‘systemic care’); taking care of impaired adult or elderly PWH (i.e. ‘transition care’); coping with child or adolescent PWH (i.e. ‘transition care’). Such emerging issues should be developed further as they offer the opportunity to clarify and expand the notion of haemophilia CC, by increasing its coherence and accuracy. Achieving a definition of CC that is both inclusive and accurate has important implications for everyday medical practices. On the one hand, it permits formulating practical recommendations and proper indicators for evaluating the effectiveness of healthcare programmes. On the other hand, it permits better comprehending the options available and stimulating developments in research projects and interventions.

In this regard, starting from these closing remarks, a research project funded by the HERO Research Grant was completed.² This project aimed to integrate the

forementioned observations, introducing a broader psychosocial perspective in the notion of CC. The project is based on the idea that each of the five themes identified by Palareti et al. [74] and Poti et al. [75] represents a subject where the use of different perspectives (HCPs, PWH, families and patients' associations) can lead to the identification of relevant cross-professional and non-technical skills that support haemophilia CC in both its assumptions [cf. also 76–78]. Asking all the actors and stakeholders involved to express their viewpoints and needs is a crucial point for improving the coordination of the care provided: indeed, in line with previous research, this process helps to 'understand sense-making processes and how roles are redefined and negotiated' [61, p. 38].

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