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ORIGINAL ARTICLE

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The psychosocial impact of haemophilia from patients' and caregivers' point of view: The results of an Italian survey

Arianna Fornari[1,2] | Ippazio Cosimo Antonazzo[1] | Angiola Rocino[3] | Daniele Preti[4] | Anna Fragomeno[4] | Francesco Cucuzza[4] | Nicola Ceresi[4] | Cristina Santoro[5] | Antonietta Ferretti[5,6] | Rita Facchetti[1] | Paolo Cozzolino[7] | Chiara Biasoli[8] | Cristina Cassone[4] | Antonio Coppola[9] | Paolo Angelo Cortesi[1] | Lorenzo Giovanni Mantovani[1,7]

[1]Research Centre on Public Health (CESP), University of Milano-Bicocca, Monza, Italy

[2]Neurology, Public Health, Disability Unit, Fondazione IRCCS Istituto Neurologico Carlo Besta, Milan, Italy

[3]UOC di Ematologia-Centro Emofilia e Trombosi, Ospedale del Mare-ASL NA1, Napoli, Italy

[4]Italian Federation of Haemophilia Associations (FedEmo), Rome, Italy

[5]Haematology, University Hospital Policlinico Umberto I, Rome, Italy

[6]Haemorrhagic and Thrombotic Diseases Service, Area of Haematology, Fondazione Policlinico Universitario 'A. Gemelli', IRCCS, Rome, Italy

[7]IRCCS Multimedica, Sesto San Giovanni, Italy

[8]Haemophilia Centre and Transfusion Department, Bufalini Hospital, Cesena, Italy

[9]Regional Reference Centre for Inherited Bleeding Disorders, University Hospital of Parma, Parma, Italy

Correspondence
Arianna Fornari, Fondazione IRCCS Istituto Neurologico Carlo Besta, Milan, 20133, Italy.
Email: arianna.fornari@istituto-besta.it

Paolo Angelo Cortesi and Lorenzo Giovanni Mantovani are joint senior authors.

Abstract

Backgroud: A huge amount of data about psychosocial issues of people with haemophilia (PwH) are available; however, these materials are fragmentary and largely outdated, failing to reflect the impact of current treatment strategies.

Aim: Describing the influence of illness on psychosocial aspects of adult PwH (≥18 years) and caregivers of children with haemophilia (CPwH) without inhibitors, in Italy.

Methods: Surveys (for adult PwH, CPwH and haemophilia specialists) were developed by a multidisciplinary working group and conducted from November 2019 to June 2020.

Results: A total of 120 PwH without inhibitors and 79 CPwH completed the survey. Adult patients reported a significant impairment in many psychosocial aspects, including working activities, relations with family members and social relations. Caregivers generally reported better scores in all aspects of the survey. Mobility, Pain and Mental health domains of EQ-5D were the most frequently impaired in both patients and caregivers, reducing the perceived quality of life. Genetic counselling was an important issue, 53% of CPwH declaring unawareness of their carrier status, as well as the psychological support offered by the reference center, 67.0% of respondents reporting that no psychological support was provided at the time of diagnosis communication.

Conclusion: This study provides information about PwH's and CPwH's point of view in the current scenario of continuous innovations in haemophilia treatment and management furthermore, updated insights on psychosocial problems faced by patients and caregivers are reported.

KEYWORDS
burden, caregivers, daily activities, haemophilia, psychosocial issues, quality of life, survey

1 | INTRODUCTION

Haemophilia is an inherited bleeding disorder caused by the deficiency of clotting factor VIII (FVIII; haemophilia A, HA) or Factor IX (FIX; Haempohilia B, HB).[1] Beyond the medical condition, individuals with a chronic disease such as haemophilia can commonly face several social, psychological and economic challenges. The social and psychological aspects can be summarized under the word 'Psychosocial', an umbrella term that unites both aspects of behaviour.[2] Psychosocial support is an important part of comprehensive care for people with haemophilia (PwH), helping to adapt and cope with a new reality, given that the medical treatment alone does not automatically translate into better quality of life.[3] Recently, the World Federation of Hemophilia (WFH) published an updated guidance on haemophilia management in which health professionals were recommended to involve patients in decision-making process, make a psychosocial assessment and give support as needed.[4] A previous survey investigated the situation about haemophilia care in Europe and showed that 58% of countries surveyed did not offer psychosocial support, highlighting an important lack.[5] Psychosocial interventions, when products for treatment, in particular for regular prophylaxis of bleeding and the related joint deterioration (currently the standard of care for PwH with severe disease or phenotype[4]) are available, are essential to optimize outcomes and improve the quality of life. In countries in which the treatment is not available or easy to access, psychosocial support plays an important role helping to activate coping strategies and to minimize the impact of the disease and related disabilities.[2,3] Several studies provided information about living with haemophilia and the issues related to disease[1,6,7–9] but, unfortunately, these studies were conducted in the first decade of 2000s, therefore the reported data does not reflect the current evolution of clinical scenario.[1,6,7–9] Indeed, in the last years, new treatment options and management approaches have been adopted, including diffusion of prophylaxis at all ages and comprehensive care and the availability of products reducing treatment burden and facilitating adherence (FVIII and FIX concentrates with extended half-life, EHL; the first non-replacement agent, the FVIII-mimetic bispecific monoclonal antibody emicizumab, administered s.c).[4] These innovations enabled to address clinical, pharmacological, genetic and psychological issues. To ensure continuing improvements in haemophilia care it is important to recognize how these changes can impact daily life of PwH and caregivers.

The aim of this study is to provide an updated picture of the impact of haemophilia in Italy, through surveys carried out in adult PwH and caregivers of children with haemophilia (CPwH) without inhibitors and physicians of specialist centres. A previous paper, recently published by Cortesi and collegues, reported the results of these surveys about the perceived disease control, treatment approaches, treatment satisfaction, and access to care in Italy.[10] The present paper focuses on the psychosocial aspects including quality of life, working activities, sport activities, social relations, and parenting of PwH and CPwH.

2 | MATERIALS AND METHODS

2.1 | Recruitment and study design

Methods for the present study have been previously described.[10] Briefly, two web-based questionnaires (one for adult patients and one for parents/caregivers) and an electronic questionnaire (for clinicians) were designed. The following domains were explored: 1. Demographic; 2. Disease characteristics and symptoms, 3. Disease control, 4. Therapeutic approach and treatment satisfaction, 5. Access to care, 6. Quality of life, 7. Working activities, 8. Sport activities, 9. Social relations and 10. Parenting.

Results related to the domain 1—5 have been described in a previuos publication.[10] Our paper focused on domains from 6 to 10. Participants were recruited through online invitations, sent by the Federation of Italian Associations of Haemophilia patients (FedEmo) and the regional associations. The survey was conducted between November 2019 and June 2020. The study included adult (≥18 years) patients with a diagnosis of HA or HB, or caregivers of children with HA or HB (< 18 years), in both cases with or without inhibitors. Only individuals with internet access and able to understand and complete the questionnaire were selected. The survey aimed to enrol as many participants as possible and, despite the Ethics Board review and approval was not required,[9] an informed consent was requested before starting the questionnaire.

2.2 | Patient outcome assessment tools

The questionnaire was composed of more than 50 items. Specifically, it included multiple-choices and open-ended questions, rating scales and the validated health related quality of life questionnaires EQ-5D-5L and EQ-5D-Y (EuroQoLGroup) with visual analogue scale (VAS). EQ-5D-5L measures current overall health status and comprises five dimensions: 'Mobility', 'Self-care', 'Usual activities', 'Pain/discomfort' and 'Anxiety/Depression'; each dimensions have five levels that range from 'no problems' to 'extreme problems'. The patients should indicate his/her health state in the most appropriate box. The EQ-VAS records the patient's self-rated health on a vertical visual analogue scale in which participants should indicate their current health status, ranging from the 'worst health you can imagine' (low scores) to 'the best health you can imagine' (high scores).[11]

2.3 | Data analysis

Descriptive analysis was conducted among PwH and CPwH without inhibitors stratified by subgroups (e.g. haemophilia type, severity of illness, prophylaxis or on-demand treatment and, among those on prophylaxis, standard vs. extended half-life concentrate treatment). Due to the highly different impact of the disease and the lower number of data collected, similarly with the previous study, inhibitor PwH and CPwH were not included in this analysis. Results were reported by

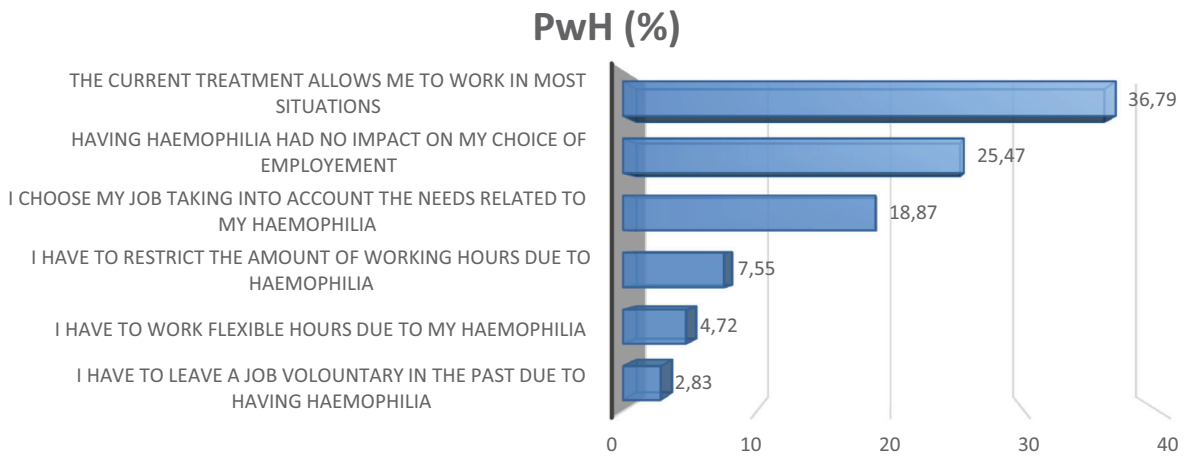

FIGURE 1 Working life of adult patients [all treatment regimens]. Legend: PWH, adult patients with haemophilia; All types of treatment, Prophylaxis or On-demand treatment.

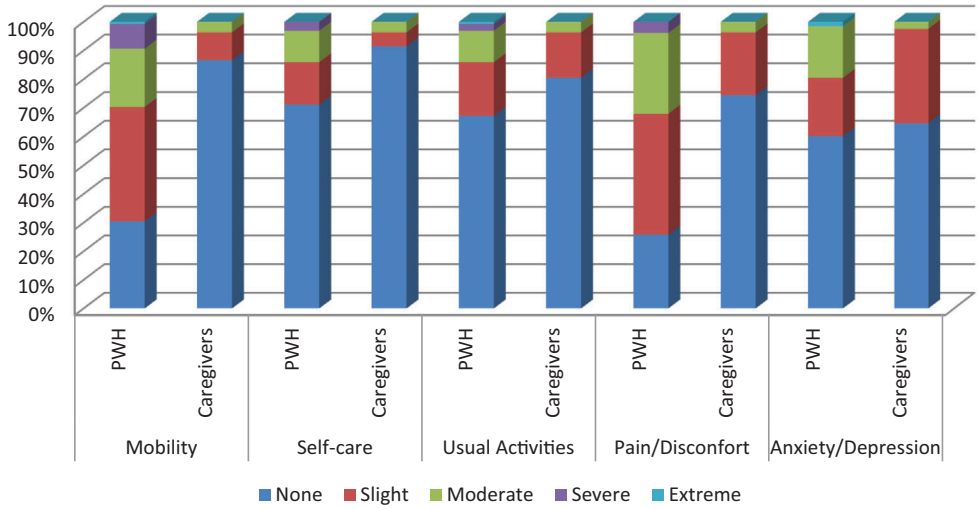

FIGURE 2 Patients' and caregivers' quality of life results of EQ-5D-5L and EQ-5D-Y. PwH, adult patients with haemophilia; Caregivers, parent/caregiver of children with haemophilia. None = no impairment; Slight = low impairment; Moderate = moderate impairment; Severe = severe impairment; Extreme = extreme impairment.

frequency tables and, where useful, presented graphically. The goal of analysis reported here was to identify possible trends across the different groups.

3 | RESULTS

The descriptive analysis was performed on 144 (60.5%) PwH and 94 (39.4%) CPwH without inhibitors. For all participants, socio-demographic and disease characteristics were reported stratifying by severity of illness and type of haemophilia (Table 1). The mean age (SD) of PwH was 45.9 (15.4), while the mean age of children with haemopylia was 11.7 (7.14). The majority of both adults and children had HA (81% of adults and 78% of children, respectively). Additionally, a significant proportion of participants had severe disease, with 66.6% of adults and 74.6% of children falling into this category. Regarding treatment, the majority of patients received regular prophylaxis (70% of PwH and 85% of children) the remaining were treated on demand than on demand (28% PwH; 15.1% children). The most reported comorbidity was hepatitis B or C infection (50%), followed by Human Immunodeficiency Virus infection (32%), cardiovascular disease (38%) and psychiatric disorder (18%). Cardiovascular diseases were present in few children (2.5%), while no CPwH reported infectious disease or mental disorders. Approximately half of PwH participants were married and more than 50% had completed secondary education (upper secondary school). Most of respondents were resident in the North of Italy (Table 1).

TABLE 1 Characteristics of adult patients (PwH) and caregivers of children with severe haemophilia (CPwH) without inhibitors.

	PwH			CPwH		
	HA/HB (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	HA/HB (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)
Age, years, mean (SD)	45.99 (15.49)	44.85 (13.86)	47.07 (16.59)	11.7 (7.14)	11.37 (6.09)	9.08 (6.56)
Weight, kg, mean (SD)	77.23 (14.44)	77.52 (16.52)	77.57 (8.4)	45.29 (23.88)	44.17 (23.95)	37.46 (25.73)
Height, cm, mean (SD)	175.26 (7.89)	175.8 (8.87)	174.57 (6.93	147.07 (33.19)	144.75 (34.2)	134.85 (37.06)
Marital status—*N* (%)						
Single	36 (30)	17 (25.76)	4 (28.57)	–	–	–
Common-low partner	16 (13.33)	9 (13.64)	4 (28.57)	–	–	–
Married	59 (49.17)	34 (51.52)	5 (35.71)	–	–	–
Separated/divorced	5 (4.17)	4 (6.06)	0 (0)	–	–	–
Widow	0 (0)	0 (0)	0 (0)	–	–	–
Prefer not to answer	4 (3.33)	2 (3.03)	1 (7.14)	–	–	–
Haemophilia type - *N* (%)						
A	96 (80.67)	66 (100)	0 (0)	62 (78.48)	46 (100)	0 (0)
B	23 (19.33)	0 (0)	14 (100)	17 (21.52)	0 (0)	13 (100)
Haemophilia severity - *N* (%)						
Severe	80 (66.67)	66 (100)	14 (100)	59 (74.68)	46 (100)	13 (100)
Moderate	18 (15)	0 (0)	0 (0)	13 (16.46)	0 (0)	0 (0)
Mild	22 (18.33)	0 (0)	0 (0)	7 (8.86)	0 (0)	0 (0)
Treatment regimens N (%)						
On-demand	34 (28.8)	5 (7.57)	3 (21.4)	12 (15.1)	1 (2.17)	0 (0)
Prophylaxis	84 (71.1)	60 (92.3)	11 (78.5)	67 (84.8)	45 (97.8)	13 (100)
Comorbidities—*N* (%)						
Hepatitis	25 (50)	16 (50)	2 (40)	0 (0)	0 (0)	0 (0)
HIV infection	16 (32)	12 (37.50)	2 (40)	0 (0)	0 (0)	0 (0)
Psychiatric disorders[a]	9 (18)	6 (18.75)	1 (20)	0 (0)	0 (0)	0 (0)
Cardiovascular diseases	19 (38)	11 (34.38)	1 (20)	2 (2.53)	1 (2.17)	1 (7.69)
Others	21 (42)	14 (43.75)	2 (40)	12 (15.18)	5 (10.86)	3 (23.07)
Education—*N* (%)						
None/Primary school	3 (2.5)	1 (1.52)	1 (7.14)	–	–	–
Lower secondary school	15 (12.5)	5 (7.58)	3 (21.43)	–	–	–
Upper secondary school	68 (56.67)	39 (59.09)	5 (35.71)	–	–	–
University degree	29 (24.17)	18 (27.27)	4 (28.57)	–	–	–
Postgraduate degree	4 (3.33)	2 (3.03)	1 (7.14)	–	–	–
Prefer no answer	1 (.83)	1 (1.52)	0 (0)	–	–	–
Residence—*N* (%)						
North of Italy	53 (44.16)	24 (36.36)	7 (50)	33 (41.77)	21 (45.65)	6 (46.14)
Centrer of Italy	33 (27.50)	17 (25.75)	5 (35.71)	12 (15.18)	8 (17.39)	0 (0)
South of Italy	34 (28.33)	25 (37.87)	2 (14.28)	34 (43.03)	17 (36.95)	7 (53.84)

Abbreviations: CPwH, caregivers of minor patients with haemophilia; HA, haemophilia A; HB, haemophilia B; HIV, human immunodeficiency virus.;PwH, adult patients with haemophilia.

[a]Anxiety, depression, insomnia, chronic fatigue and acute stress.

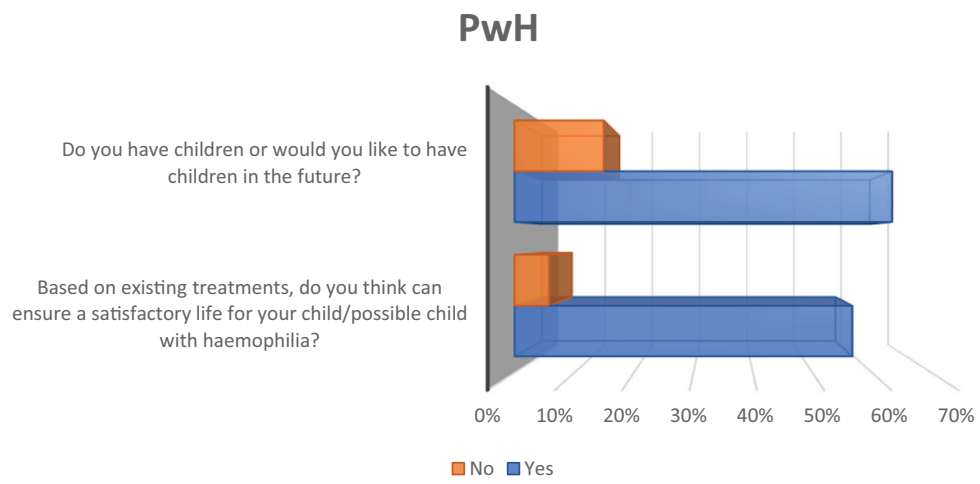

FIGURE 3 Parenting pathway of adult patients with severe haemophilia. PWH, adult patients with haemophilia; CPwH, parent/caregiver of children with haemophilia.

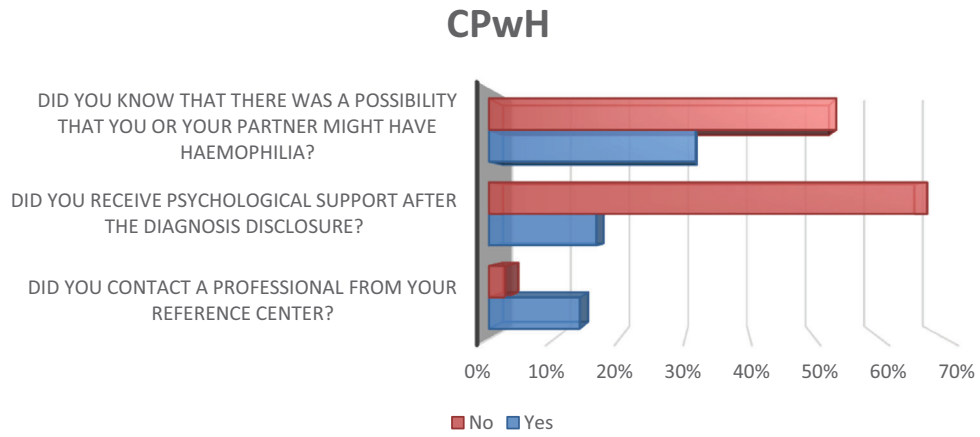

FIGURE 4 Parenting pathway of caregivers of children with severe haemophilia. PWH, adult patients with haemophilia; CPwH, parent/caregiver of children with haemophilia.

3.1 | Working and scholastic activities

Almost half of PWH (48.3%) have a full-time work and a small percentage (15%) is unemployed, however 38.8% of the latter reported that they do not have a job due to heamophilia. Approximately one-third of PwH received disability benefits, with higher rates observed in severe HA (42.4%) patients compared to severe HB (21.4%). About one out of five (18.8%) of PwH declared that the job was chosen considering the needs related to haemophilia and few patients reported modifications of working hours (12.2%) or voluntary resignation (2.8%) due to the illness (Figure 1). Most children with hemophilia attend the middle or high school and 18.9% of CPwH reported that the choice of school was conditioned by illness, especially for HB patients (31%). Caregivers' satisfactions with the management of haemophilia management by school setting was high, reaching (67.0%) for both type of haemophilia (Table 2). Regarding the impact on work and school, the number of days lost at school or at work due to haemophilia was low. However, approximately 40% of adult patients reported a small or moderate impact on their work due to the illness and 10% considered to be high. On the other hand, almost half of CPwH did not report any negative impact on school activities (Table 2).

3.2 | Sport and leisure activities

Physical activities were practiced by 42.5% of adult PwH and 64.5% of children; higher rates were found in adult patients with severe HB (57.1%) and children with severe HA (63%). Overall, more than half of PwH and children engaged in sport activities one or two time a week. Swimming was the most practiced physical activity. Many adult patients reported regular participation in walking, jogging and football. A high percentage of PwH (61.6%) and children (40.5%) expressed the desidere to engage in activities that were restricted due to their

TABLE 2 Working and scholastic activities by adult patients and caregivers with severe haemophilia.

	PwH			CPwH			*p* (PwH vs. CPwH)		
	All (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	ALL (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)	HA/HB	Severe HA	Severe HB
Main working activities—*N* (%)									
Full-time work	58 (48.33)	34 (51.51)	8 (57.14)	–	–	–	–	–	–
Housewife/Homemaker	1 (.83)	1 (1.51)	0 (0)	–	–	–	–	–	–
Retired	11 (9.16)	4 (6.06)	1 (7.14)	–	–	–	–	–	–
Part-time word	10 (8.33)	8 (12.12)	0 (0)	–	–	–	–	–	–
Student	9 (7.5)	4 (6.06)	2 (14.28)	–	–	–	–	–	–
Unemployed*	18 (15)	8 (12.12)	2 (14.28)	–	–	–	–	–	–
School class—*N* (%)									
Kindergarden	–	–	–	8 (10.12)	3 (6.52)	3 (23.07)	–	–	–
Primary school	–	–	–	14 (17.72)	9 (19.56)	3 (23.07)	–	–	–
Middle school	–	–	–	20 (25.31)	10 (21.73)	2 (15.38)	–	–	–
High school	–	–	–	21 (26.58)	13 (28.26)	3 (23.07)	–	–	–
*Unemployed due to haemophilia									
Yes	7 (38.89)	4 (50)	1 (50)	–	–	–	–	–	–
Disability allowance due to haemophilia									
Yes	36 (33.96)	28 (42.42)	3 (21.42)	–	–	–	–	–	–
School choice conditioned by hHaemophilia									
Yes	–	–	–	15 (18.98)	9 (19.56)	4 (30.76)			
Impact of haemophilia on employment/scholastic activities—*N* (%)							.008	.0108	.4189
A high negative impact	12 (10)	5 (7.57)	1 (7.14)	1 (1.26)	1 (2.17)	0 (0)	–	–	–
A moderate negative impact	25 (20.83)	17 (25.75)	2 (14.28)	7 (8.86)	2 (4.34)	1 (7.69)	–	–	–
A small negative impact	27 (22.5)	11 (16.66)	5 (35.71)	21 (26.58)	11 (23.91)	2 (15.38)	–	–	–
No negative impact	42 (35)	25 (37.87)	5 (35.71)	37 (46.83)	23 (50)	8 (61.53)	–	–	–
Satisfaction of haemophilia management by school									
Yes	–	–	–	53 (67.08)	31 (67.39)	8 (61.53)			
N of day loss at school or at work due to Haemophilia—*N* (%)							.8626	.7454	.1924

(Continues)

TABLE 2 (Continued)

	PwH			CPwH			*p* (PwH vs. CPwH)		
	All (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	ALL (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)	HA/HB	Severe HA	Severe HB
0	37 (30.83)	15 (22.72)	9 (64.28)	29 (36.70)	17 (36.95)	5 (38.46)	–	–	–
1–5	27 (22.5)	20 (30.30)	1 (7.14)	25 (31.64)	15 (32.60)	3 (23.07)	–	–	–
6–10	6 (5)	3 (4.54)	0 (0)	5 (6.32)	1 (2.17)	1 (7.69)	–	–	–
11–15	5 (4.16)	3 (4.54)	0 (0)	3 (3.79)	2 (4.34)	(0)	–	–	–
16–20	3 (2.5)	2 (3.03)	0 (0)	1 (1.26)	1 (2.17)	(0)	–	–	–
>20	4 (3.33)	2 (3.03)	0 (0)	1 (1.26)	0 (0)	1 (7.69)	–	–	–

Abbreviations: CPwH, caregivers of minor patients with haemophilia; HA, haemophilia A; HB, haemophilia B.;PwH, adult patients with haemophilia.

disease. Some PwH refrained from participating in certain activities due to concerns about treatment coverage, accounting for 21.6% of cases.

CPwH reported that sports were not practiced due to children's or caregivers' conditioning by illness in 26.5% and 31.2% of cases, respectively. As regards the leisure activities, such as travel or class trip, approximately 22% of caregivers and 28% of adult patients reported a moderate or high negative impact of haemophilia on such activities (Table 3).

3.3 | Quality of life

Overall, the majority of PwH and CPwH reported no problems in most domains of EQ-5D. More than half of participants reported no issues with taking in take care of themselves (PwH - 68%; caregivers - 85%), carrying out their usual activities (PwH - 64%; caregivers - 76%) and experienced no problems related to Anxiety or Depression (PwH - 56%; caregivers - 58%). However, 36% of PwH reported slight problems in 'Mobility' and 48% in 'Pain/Discomfort' domains, at variance with most caregivers, who reported no problems in these domains (Figure 2). The EQ-VAS showed a higher perceived health status among caregivers (mean 83.39) compared to PwH (mean 73.34). When analysing the results in adult patients and caregivers, stratified by treatment regimen, significant impairments were observed (Supplementary figure 1 and 2). PwH treatment reported the most critical issues in 'Mobility' and 'Pain/Discomfort' domains, with over 50% reporting mild to moderate problems. Moreover, more than half of those treated on demand reported mild or moderate problems in the 'Depression/Anxiety' domain. Overall, CPwH reported no difficulties in most domains, regardless of treatment regimens. However, caregivers of children receiving regular prophylaxis reported mild problems in 'Anxiety or Depression' (35%) and 'Pain/Discomfort' (21%) domains. Among CPwH treated on-demand, 33% reported mild problems in the 'Pain/Discomfort' domain (Supplementary figure 1 and 2).

3.4 | Social participation

Regarding social relations, most patients reported only disclosing their diagnosis to a small group of people. PwH preferred to share their diagnosis with partner (68%), relatives (65%) and friends (51%). Similarly, CPwH preferred to share the diagnosis with relatives (86%), friends (75%) and siblings (67%). However, CPwH also disclosed the diagnosis to their children's teachers (75%) and schoolmates (57%). Negative impact of haemophilia on establishing social relations was reported by 33% of adult patients and 13% of caregivers.

No negative impact on partner's life (58%) was reported by PwH, while a low (29%) or moderate (20%) impact of illness was reported on family members. Generally, CPwH expressed satisfaction with the support received from their partner (72%), family members (57%) and other contacts related to their children (56%) (Table 4). In Supplementary figure 3, the results stratified by treatment regimens showed that

TABLE 3 Sport and leisure activities in patients and caregivers of children with of haemophilia.

	PwH			CPwH			*p* (PwH vs. CPwH)		
	ALL (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	ALL (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)	HA/HB	Severe HA	Severe HB
Physical activities									
Yes	51 (42.5)	22 (33.33)	8 (57.14)	51 (64.55)	29 (63.04)	7 (53.84)	.0004	.0002	1.0000
Frequency of physical activities									
Every day	4 (7.84)	3 (13.64)	1 (12.5)	3 (5.88)	2 (6.89)	0 (0)	–	–	–
Twice a week or more	18 (35.29)	9 (40.91)	1 (12.5)	23 (45.1)	14 (48.27)	2 (28.57)	–	–	–
One or two times a week	25 (49.02)	9 (40.91)	4 (50)	21 (41.18)	12 (41.38)	3 (42.86)	–	–	–
Less than once a week	4 (7.84)	1 (4.55)	2 (25)	4 (7.84)	1 (3.45)	2 (28.57)	–	–	–
Type of physical activities									
Swimming	24 (47.06)	9 (40.91)	5 (62.5)	25 (49.01)	15 (51.72)	2 (28.57)	–	–	–
Fishing	1 (1.96)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	–	–	–
Soccer	5 (9.8)	2 (9.09)	1 (12.5)	7 (13.72)	6 (20.69)	1 (14.28)	–	–	–
Horse riding	1 (1.96)	1 (4.55)	0 (0)	0 (0)	0 (0)	0 (0)	–	–	–
Cycling	6 (11.76)	2 (9.09)	2 (25)	2 (3.92)	1 (3.45)	0 (0)	–	–	–
Basketball	1 (1.96)	0 (0)	0 (0)	4 (7.84)	2 (6.9)	1 (14.28)	–	–	–
Tennis	2 (3.92)	1 (4.55)	1 (12.5)	6 (11.76)	4 (13.79)	1 (14.28)	–	–	–
Dance	1 (1.96)	1 (4.55)	0 (0)	0 (0)	0 (0)	0 (0)	–	–	–
Volleyball	0 (0)	0 (0)	0 (0)	2 (3.92)	1 (3.45)	0 (0)	–	–	–
Yoga	1 (1.96)	1 (4.55)	0 (0)	0 (0)	0 (0)	0 (0)	–	–	–
Running	7 (13.73)	2 (9.09)	3 (37.5)	2 (3.92)	0 (0)	2 (28.57)	–	–	–
Regular walking	23 (45.1)	10 (45.45)	5 (62.5)	7 (13.72)	6 (20.69)	1 (14.28)	–	–	–
Travel or class trip									
A high impact	8 (6.66)	7 (10.60)	0 (0)	6 (7.59)	2 (4.34)	2 (15.38)	–	–	–
A moderate negative impact	26 (21.66)	11 (16.66)	7 (50)	11 (13.92)	5 (10.86)	2 (15.38)	–	–	–
A very low negative impact	37 (30.83)	20 (30.3)	2 (14.28)	22 (27.84)	14 (30.43)	3 (23.07)	–	–	–

(Continues)

TABLE 3 (Continued)

	PwH			CPwH			*p* (PwH vs. CPwH)		
	ALL (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	ALL (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)	HA/HB	Severe HA	Severe HB
No negative impact	35 (29.16)	20 (30.3)	4 (28.57)	28 (35.44)	16 (34.78)	5 (38.46)	-	-	-
Activities not practiced, but desired									
Yes	74 (61.66)	44 (66.66)	10 (71.42)	32 (40.50)	16 (34.78)	7 (53.84)	.0020	.0005	.4110
Activities not practiced due to feel conditioned by coverage of treatment									
Yes	16 (21.62)	11 (25)	0 (0)	-	-	-	-	-	-
Activities not practiced by children due to caregivers' conditioning caused by haemophilia									
Yes	-	-	-	10 (31.25)	6 (37.5)	1 (14.28)	-	-	-
Activities not practiced by children due to conditioning by haemophilia									
Yes	-	-	-	21 (26.58)	11 (68.75)	1 (14.28)	-	-	-

Abbreviations: CPwH, caregivers of minor patients with haemophilia; HA, haemophilia A; HB, haemophilia B.;PwH, adult patients with haemophilia.

40% of adult patients treated on demand reported a mild impact of illness on their partners'lives and other family members. Adult PwH on regular prophylaxis reported a mild impact on family members in 30% of cases and no negative impact on their partners' life in 73% of cases.

3.5 | Parenting pathway

Approximately 63.0% of adult patients reported having children or expressed a desire to have children in the future. Among them, 57.0% believe that the available treatments could ensure a satisfactory life for their children in the future. Around half of the mothers (53%) declared that they were not aware about their haemophilia carrier status, and 67.0% did not receive psychological support when the diagnosis was communicated to them. However, some adult PwH reported receiving support from a mental health professional, although only 38.0% of them provided by the haemophilia treatment center (Figures 3 and 4).

3.6 | Type of prophylaxis

As shown in in Table 1 in the supplementary material, among PwH, 42 patients received prophylaxis with extended half-life concentrates while 40 were on prophylaxis with standard half-life products. The majority of patients in both groups were full-time employed, 55% and 61%, respectively.The results did not show significant difference between the studied groups in terms of the impact of prophylaxis type on work status. Among CPwH, over 80% of subjects in both groups reported that the disease has a small or no negative impact on their scholastic activities. Once again, the results were not statistically significant. Additionally, in both groups, patients lost from 0 to 5 work days due to the disease, with no significant difference observed. PwH on extended half-life prophylaxis were more likely to be engaged in physical activities ($p = .03$). Among CPwH, five individuals using extended half-life prophylaxis reported a high impact on travel or class trips (Table 2 supplemental material). As reported in Table 3 in the supplemental material, PwH individuals under standard prophylaxis reported a negative impact of the disease on their social relations compared to those under extended half-life prophylaxis. As reported in Table 4 in the supplemental material, the use of the different types of prophylaxis regimens did not show significant difference between studied groups across the different EQ-5D-5L domains except for 'Depression/Anxiety'. Among PwH, those treated with extended half-life prophylaxis reported higher frequency of the absence of anxiety compared to the other regimen.

4 | DISCUSSION

This study provides an updated picture of the impact of haemophilia on psychosocial aspects (quality of life, working activities, sports, social relations and parenting), investigated in a recent Italian survey. The results showed that many psychosocial factors, affected by

TABLE 4 Social participation of adult patients and caregivers of children with severe haemophilia.

	PwH			CPwH			*p* (PwH vs. CPwH)		
	ALL (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	HA/HB no inhibitors (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)	HA/HB	Severe HA	Severe HB
Diagnosis disclosure									
Wife/husband or partner	82 (68.33)	46 (69.69)	11 (78.57)	–	–	–	–	–	–
Kids	37 (30.83)	19 (15.83)	5 (35.71)	–	–	–	–	–	–
Relatives	78 (65)	44 (36.66)	10 (71.42)	68 (86.07)	39 (84.78)	11 (84.61)	–	–	–
Friends	61 (50.83)	34 (51.51)	8 (57.14)	59 (74.68)	34 (73.91)	11 (84.61)	–	–	–
Colleagues/schoolmate	43 (35.83)	25 (37.87)	6 (42.85)	45 (56.96)	27 (58.69)	7 (53.84)	–	–	–
Other contacts	15 (12.5)	4 (6.06)	3 (21.42)	17 (21.51)	13 (28.26)	4 (30.76)	–	–	–
Brothers/Sisters	–	–	–	53 (67.08)	28 (60.86)	10 (76.92)	–	–	–
Teachers	–	–	–	59 (74.68)	34 (73.91)	10 (76.92)	–	–	–
Negative impact of haemophilia on establish social relations									
Yes	40 (33.33)	22 (33.33)	2 (14.28)	10 (12.65)	6 (13.04)	1 (7.69)	.0199	.0196	1.0000
Impact of haemophilia on partner									
Very high	2 (1.66)	1 (1.51)	0 (0)	–	–	–	–	–	–
Moderate	12 (10)	6 (9.09)	1 (7.14)	–	–	–	–	–	–
Very low	19 (15.83)	9 (13.63)	3 (21.42)	–	–	–	–	–	–
No negative impact	69 (57.5)	39 (59.09)	8 (57.14)	–	–	–	–	–	–
Impact of haemophilia on family members									
Very high	8 (6.66)	6 (9.09)	0 (0)	–	–	–	–	–	–
Moderate	24 (20)	12 (18.18)	4 (28.57)	–	–	–	–	–	–
Very low	35 (29.16)	16 (24.24)	6 (42.85)	–	–	–	–	–	–
No negative impact	38 (31.66)	23 (34.84)	3 (21.42)	–	–	–	–	–	–
Satisfaction of partner's support									
Very satisfied	–	–	–	57 (72.15)	33 (71.73)	9 (69.23)	–	–	–
Low satisfied	–	–	–	1 (1.26)	1 (2.17)	0 (0)	–	–	–
Low dissatisfied	–	–	–	6 (7.59)	4 (8.69)	1 (7.69)	–	–	–
Very dissatisfied	–	–	–	2 (2.53)	1 (2.17)	1 (7.69)	–	–	–
Prefer no answer				3 (3.79)	0 (0)	1 (7.69)	–	–	–

(Continues)

TABLE 4 (Continued)

	PwH			CPwH			*p* (PwH vs. CPwH)		
	ALL (*N* = 120)	Severe HA (*N* = 66)	Severe HB (*N* = 14)	HA/HB no inhibitors (*N* = 79)	Severe HA (*N* = 46)	Severe HB (*N* = 13)	HA/HB	Severe HA	Severe HB
Satisfaction of family members' support									
Very satisfied	–	–	–	45 (56.96)	25 (54.34)	6 (46.15)	–	–	–
Low satisfied	–	–	–	8 (10.12)	7 (15.21)	0 (0)	–	–	–
Low dissatisfied	–	–	–	6 (7.59)	4 (8.69)	2 (15.38)	–	–	–
Very dissatisfied	–	–	–	4 (5.06)	3 (6.52)	1 (7.69)	–	–	–
Prefer no answers	–	–	–	6 (7.59)	0 (0)	3 (23.07)	–	–	–
Satisfaction of support by other contacts of their children									
Very satisfied	–	–	–	44 (55.69)	24 (52.17)	6 (46.15)	–	–	–
Low satisfied	–	–	–	10 (12.65)	6 (13.04)	1 (7.69)	–	–	–
Low dissatisfied	–	–	–	7 (8.86)	6 (13.04)	1 (7.69)	–	–	–
Very dissatisfied	–	–	–	2 (2.53)	1 (2.17)	1 (7.69)	–	–	–
Prefer no answere	–	–	–	5 (6.32)	1 (2.17)	3 (23.07)	–	–	–

Abbreviations: CPwH, caregivers of minor patients with haemophilia; HA, haemophilia A; HB, haemophilia B;PwH, adult patients with haemophilia.

haemophilia, contribute to influence the patients' daily life. In our study the median age of PwH population was 46 years and most of the respondents had moderate or severe haemophilia. Educational level was high for both PwH and children, in line with previous rersearch.[12,13] Olny a small number of PWH do not have a job but, among these, a relevant propotion mentions illness as the cause of unemployment and received disability benefits. This result is in line with other international studies that report an important impact of illness on working area. It is possible that the severe arthropathy developed by patients, born before the '90, or the restriction in their job performance may contribute to limit the access to the working palce.[2,13] Some CPwH reported the influence of disease on school choice, while scholastic activities were not negatively affected and the satisfaction with management of haemophilia was high.

Despite the PWH practice less sport activities than children, either report engaging in low-risk sports in line with the recommendation by the US National Hemophilia Foundation and the World Foundation of Hemophilia.[15,16] Furthermore, physical activity is often associated with positive effects on physical well-being, on self-esteem and social interactions.[17] Swimming was the most practiced both in adults and children, but some higher risk activities were also reported, in particular soccer (by adults and children), basketball and volleyball (by children). Adults and children, even with severe haemophila, are currently able to play many hours of physical activity thanks to the available treatment regimens of regular and personalized prophylaxis[4]; indeed, some studies reported that in developing countries, where prophylaxis is unfeasible, the sport participation remains poor.[18,19] PwH and children reported a strong desire to practice some physical activities not allowed due to haemophilia because both report the fear of having a new bleed. These findings are related to the higher risk of activities desired and for fear of injuries and complications.[20] In our survey for few patients, travel represents an issue. Generally, the frequently infusion and storage condition of treatment might represent an issue for patients.[21] However, it should be noted that the new therapies require less infusion and therefore have lower impact on patietns ability to travel. Furthermore, patients on prophylaxis with Emicizumab might take more advantage by this therapy due to peculiar administration schedule (long time intervals: 1, 2 or 4 weeks).[4] New studies should be performed to deeply investigate how patients manage their therapy during a travel and potential issues related to travel.

The daily challenges faced by people with haemophilia and their caregivers can have major repercussions on well-being.[5] Previous research reported that the participants generally had a lower QoL compared with people who do not have haemophilia.[22,23] In our study, the EQ-5D showed that most adult patients with severe haemophilia reported slight problems in Mobility and Pain/Discomfort domains and the severity of impairment increased when patients were stratified by treatment regimen. Especially for patients treated 'on demand', the problems of Anxiety or Depression add to the Mobility and Pain difficulties. Previous studies provided similar results, highlighting how in moderate or severe patients the reduced mobility is often associated with perceived pain, discomfort, and mental health problems.[1,24,25] Some authors reported that lowest scores in PwH at the EQ-5D questionnaire were due to the presence of comorbidity, like HIV infection or arthropathy,[26,27] and this finding was confirmed in our survey, especially for PwH. Parents of children with haemophilia generally reported no problems in most domain of EQ-5D and only a small proportion had mild problems in Pain/Discomfort or Anxiety/Depression domains. Such overall better quality of life is likely to reflect the psycho-physical benefits of primary prophylaxis, started since the first years of life, that allow children to grow without joint problems and hyper-protection, thus with the habit to practice regularly physical activity and sports, which contribute to improve their joint health and well-being.[17] The presence of pain and psychological problems reported by some CPwH can reflect difficulties related to the age of children. Indeed, entry into adolescence can lead to increased social stigma and difficulties with adherence to treatment, worsening the perceived quality of life and health state.[28–30] Furthermore, the parents' vision may differ from children perception, falling to report the real health-related quality of life.[31]

Haemophilia can have a huge impact not only on personal life of patients but also on parents, siblings and relatives.[32–34] In our survey, most patients reported to communicate the diagnosis only with a small group of people, in line with previous studies.[32–34] A negative impact of haemophilia on family members and on establishing social relations was reported by non-negligible proportions of PwH. These findings can be explained with a gap of knowledge about haemophilia amongst general public and/or with difficulties to access to prophylactic treatment in early life, which may have contributed to create a social stigma.[3,25] Another explanation can be given by the lack of attention to psychosocial aspects within the families from the beginning of disease manifestation, which may cause a lack of 'normalization' in themselves, making difficulties their communication with the external. However, also the burden of lifelong prophylaxis can have an impact on family members and on social relations.[30,32–35]

If the treatment regimen is considered, PwH treated on demand reported lower score of satisfaction with support than those on prophylaxis regimen. Indeed, on-demand treatment is not effective in reducing the risk of bleeding and arthropathy, the major cause of morbidity in severe haemophilia, therefore possibly increasing the burden of illness not only in patients, but also in family members and partner[32–35] (Supplementary figure 3).

Data on parenting pathway showed that most CPwH were unaware of their genetic carrier condition. This finding can be due, at least in part, to the high and increasing proportion of sporadic newly diagnosed haemophilia patients.[35] However, the importance of genetic counselling was highlighted in most studies, especially for women intending to become pregnant, to make an informed decision, or for parents with a daughter.[2,35] Overall, our results showed a positive perception of parenting conditions; indeed, most PwH declares to have children or would like to have children in the future and more than half believe that available treatment could guarantee a satisfactory life for their son. In the challenging phase of the diagnosis communications most respondents report that they did not receive psychological support. Such an intervention is important for the emotional wellbeing of patients and their caregivers, not only at the moment of diagnosis disclosure but

through the entire life cycle.[36,37] In particular, mental health support aims to avoid the rejection of haemophilia, like fear of reporting bleedings and non-compliance,[3,38,39] improving health state and quality of life. A high number of programs to provide emotional support was proposed by different health professionals, however the lack of awarenees on psychosocial care and high heterogeneity of the available literature does not allow to determine the best approaches to the comprehensive patient care.[3] Haemophilia is a chronic condition affecting each person and undermining mental health, beyond the physical problems, and it is necessary, for treatment centers, to provide psychosocial care as part of an integrated multidisciplinary approach, as clearly highlighted by the recent WFH guidelines.[40]

Our study presents some limitations: first, the use of online survey may have contributed to collect the data by a single perspective, of the caregivers, falling to capture the perceptions of all family members. Second, the children's perception may differ from that of their parents, giving partial information. Third, the results can be difficult to extend to a larger population. On the other hand, our study provides detailed information about a relatively small number of individual and can be useful for capture the point of view of people with haemophilia in the current scenario of innovations in the treatment and management approaches.

5 | CONCLUSION

This study provides a recent picture of people living with haemophilia in Italy, highlighting the psychosocial aspects that are impaired and to be addressed in the comprehensive care. Our findings confirm and extend the notion that haemophilia affects many aspects of an individual's life and that psychosocial factors have a central role in quality of life, working activities, sports, travelling and interpersonal relationships of people with haemophilia and their caregivers. However, it is probably still early to detect a significant impact of innovations and long-term studies are needed. The descriptive analysis used in this study can be useful to generate hypotheses for further investigation and may help increase awareness on psychosocial issues and ensure their consideration in the global care of people with haemophilia. Finally, the information from this paper combined with those from the first paper reporting the results of this survey can provide a complete and updated picture of patients', caregivers' and clinicians' perceptions of disease control, treatment approaches, treatment satisfaction, access to care and psychosocial burden in Italy.

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Paolo Angelo Cortesi has received a research grant from Angelini, and speaking honoraria from Pfizer and Roche.

Angiola Rocino has attended Advisory Board meetings and/or received fees as speaker in meetings organized by Bayer, CSL Behring, Kedrion, Novo Nordisk, Pfizer, Roche, Shire/Takeda and Sobi.

Daniele Preti, Antonietta Ferretti, Francesco Cucuzza, Nicola Ceresi, Ippazio Cosimo Antonazzo, Rita Facchetti, Paolo Cozzolino, Cristina Cassone have nothing to declare.

Cristina Santoro has received honoraria for consulting or speaker bureau from Novo Nordisk, Roche, Takeda, Bayer, CSL Behring, Sobi.

Chiara Biasoli acted as a paid consultant to Bayer, Roche, CSL Behring and Novo Nordiskand received fees as an invited speaker by Sobi and Takeda.

Antonio Coppola received fees as a consultant or advisory board member or invited speaker by Bayer, Kedrion, Novo Nordisk, Roche, Sobi, Takeda and Werfen.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

Research ethics committee (REC) approval was not required for the preparation of this manuscript.

ORCID

Arianna Fornari https://orcid.org/0000-0003-4228-5473
Cristina Santoro https://orcid.org/0000-0002-7181-447X
Antonietta Ferretti https://orcid.org/0000-0001-7115-8743
Antonio Coppola https://orcid.org/0000-0003-3697-706X
Paolo Angelo Cortesi https://orcid.org/0000-0001-5241-4473

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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