Persons With Hemophilia of Generation Y and Their Relatives Attitudes and Expectations From Treatment

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Abstract
This multicenter cohort study aimed to determine the attitudes and expectations of persons with hemophilia of Generation Y (PwH-Y) toward hemophilia and its treatment comparatively with the opinions of their non-hemophiliac relatives. The study was representative regarding quota-control variables of hemophiliacs registered to the provincial representatives of the Hemophilia Society of Turkey in 4 geographic regions and Istanbul. Sixty-four PwH-Y (62 males) and their 56 first-degree relatives (17 males; Generation X/baby boomers) were interviewed face-to-face using mixed data collection method. "Focus Group Study" method was used for qualitative data. Treatment adherence, requirements, and social activities were questioned with a semi-structured form. Treatment adherence rate of the PwH-Y (46.2%) was lower than that perceived by their relatives (71.4%) (p ≤ 0.05). Vascular access problems were the most common reasons for non-adherence (60% in PwH-Y and 25% in relatives). Among the components the hemophiliacs and their relatives needed most, support for accessibility of drugs/treatment ranked first (41.1% and 45%, respectively), followed by emotional support (26.1% and 32.5%, respectively). For increasing treatment success in PwH-Y, treatment should be personalized and shaped based on personal requirements.

Introduction

A cohort of people born within a particular span of time and shaped by similar events, trends or developments is called as a generation. Generation Y is defined as the people born between 1981 and 1999 inclusive.[1] This approach emphasizes focusing on people according not to their birth date but to a particular span of time.[2] In health situation analysis, "age" variable is of importance in terms of sociodemographic features and habits of an individual. However, as "age" variable provides data depending on time (i.e., trend effect), it incorporates several effects/characteristics/variables such as use of technology, being concerned about climate and environment, geographical area, consumption attitudes and behaviors, inflation-income-growth, educational status, and cultural consumption habits. Defining generation based on age groups rather than age has entailed correctly establishing the age groups. In order to the above-mentioned effects to be covered by age groups, age groups should be based on "generation approach."[3] The characteristics of Generation Y include Internet and technology dependence, high ego, continuous and persistent demand, a high proclivity for quitting job, a short attention span, chronic boredom, a desire for hierarchy to be based on success rather than seniority, a tendency for globalization, being open to change, a tendency for self-management, an attitude of attaching importance to career, seeking flexibility in career, realism, and belief in (perception of) capability of doing everything.[2,3]

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Table 1. Distribution of Generations Within the General Population and Hemophilia Populations of Turkey and Within the Study Sample.

Generation (year of birth range)	General population of Turkey (%)	Hemophilia A population of Turkey (%)	Hemophilia B population of Turkey (%)	Study sample	
				PwH-Y n (%)	Relatives of PwH-Y n (%)
Silent generation (1925-1945)	3	6	6	–	–
Baby Boomers (1946-1960)	9.5	20	21	–	–
Generation X (1961-1980)	23.5	13	14	1 (1.6)	51 (91.1)
Generation Y (1981-1999)	40	47	45	60 (93.8)	5 (8.9)*
Generation Z (after 2000)	24	14	14	3 (4.7)**	–
Total	100.0	100.0	100.0	64 (100.0)	56 (100.0)

PwH-Y, persons with hemophilia of Generation Y.
*These relatives have been living with the patients for at least 15 years.
**These patients were included in the study because they were born in 2000.

Therefore, the members of Generation Y differ significantly from the members of other generations.

Based on the 2016 data from the Turkish Statistical Institute (TurkStat), nearly half of the population in Turkey is the member of Generation Y.[4,5] According to the 2014 data of the World Federation of Hemophilia (WFH) Annual Global Survey, which were released in 2015 by the WFH, revealed that nearly 45% of all hemophiliac individuals from 82 countries in the world including Turkey were the members of Generation Y.[6] Since Generation Y accounts for a substantial proportion of general population and shows different attitudes and behaviors as compared with other generations in terms of several variables, evaluation of the perception, attitudes and behaviors of persons with hemophilia of Generation Y (PwH-Y) toward hemophilia treatment and comparisons of outcomes with the opinions of individuals of different generations (such as relatives of PwH-Y) could contribute to the success of hemophilia. The present study aimed to determine the attitudes and behaviors of PwH-Y in Turkey toward hemophilia treatment and their suggestions for the solutions related to treatment problems comparatively with the opinions of their non-hemophiliac first-degree relatives and thereby to provide information for healthcare workers about the problems determined.

Materials and Methods

The present study was designed as a multicenter cohort study and conducted between November 2016 and May 2017. Turkey has 7 geographic regions based on both its ethnographic and geographical structure. In the study, of these 7 geographical regions, one central province from 4 geographical regions and Istanbul that represents the population composition of Turkey were selected, which cover 71.43% of the features and have high representation and accessibility. Ethnographic and analytical assessments were performed by gathering hemophiliacs and their relatives living in different cities of the same region together in one of the central cities of that geographical region. The study was representative at a confidence interval (CI) of 95% with respect to the quota-control variables including age and educational and socioeconomic status of hemophiliac individuals registered to the provincial representatives of the Hemophilia Society of Turkey in that region. Accordingly, the study centers were the provincial representatives of the Hemophilia Society of Turkey and the other local societies, which were identified as the sampling nodes that ensure the validity, reliability and representation of a quantitative or mixed attitude and behavior studies. These centers were selected from Istanbul, Antalya, Trabzon, Tekirdag and Konya provinces.

Patients were selected by applying "List Sampling" based on the simple random sampling method.[7] None of the patients included in the study were using any of the new treatment options of hemophilia such as emicizumab, extended half-life recombinant factor VIII (rFVIII)/IX concentrates, and fitusiran. The relatives of patients were non-hemophiliac mother, father or elder sister or brother, spouse, of grandmother/grandfather of any patient, who were at least 1 generation older than the patient and/or have been living with the patient for at least 15 years. The majority of the PwH-Y relatives were from Generation X or Baby-Boomers (Table 1). During the sample selection process, based on a high sampling ratio of nearly 10%, 64 PwH-Y and 56 first-degree relatives were randomly selected by imposing generational and regional quotas. Based both on a high sampling ratio and on selection systematic and method, a sample size of 120 was considered appropriate according to the Central Limit Theorem.

The individuals enrolled in the present study were subjected to face-to-face interview that was performed using the mixed data 2collection technique, which obtains both quantitative and qualitative data.[7] The qualitative study was performed using the "Focus Group Study" method and a total of 15 group study was performed. In the research process, a professional service was received from Dinamo Consulting Company (Toronto-

Canada, Istanbul-Turkey) for the design, implementation, and analysis. At every stage of this process, 1 biostatistician, 2 hematologists, 1 genetics specialist, 1 psychologist, and 1 nurse among the members of the Scientific Board of the Hemophilia Society of Turkey provided counseling. At the end of the interviews, a semi-structured (open-ended) questionnaire form (including 15 questionnaires) was given to each participant in the group and they were asked to fill the forms on their own. The questionnaires in the form included questions about treatment adherence, primary requirements, and daily social activities including social media usage. The patients and their relatives were asked to specify their level of agreement (agree-disagree) independently for each statement in the questionnaire on a 10-point equal interval scale. In addition to the questions about patients' adherence to treatment (received treatments, method, dosage, and frequency of the treatments) in the semi-structured form, patient dairies provided for the patients from their treatment centers or units where they were followed-up were also used to assess patients' adherence to treatment.

The present study and the semi-structured questionnaire form used in the study were approved by the Ethics Committee of Istanbul Arel University (decision number: 2017/02, date: 24/3/2017). All patients and their relatives provided informed consent for participation in the study.

Statistical Analysis

Data analyses were performed using the IBM SPSS Statistics for Windows, Version 22.0 (IBM Corp. Armonk, NY, USA). The sample size ratios for the patient and relative groups are %53.3 and %46.7, respectively. These ratios enable statistical comparisons of samples.[7] The obtained data were analyzed using t-test, which is used for assessing the significance of difference between two parameters, at a significance level of $p \leq 0.05$.

Results

The present study included 64 PwH-Y and their 56 non-hemophiliac first-degree relatives. The distribution of generations within the general population of Turkey, within the hemophilia (hemophilia A and B) population of Turkey, and within the study sample is presented in Table 1. The general characteristics of the PwH-Y and their non-hemophiliac first-degree relatives are summarized in Table 2. Of the PwH-Y included in the study (n = 64), 97% (n = 62) had hemophilia A/hemophilia B. All 56 PwH-Y having severe type hemophilia (including 3 patients with inhibitors) were using a prophylactic factor. The patients with non-severe hemophilia A (n = 3) and hemophilia B (n = 3) and 1 patient with von Willebrand's disease (vWD) type 2 and 1 patient with Factor X deficiency were receiving on-demand therapy.

In the study, 87.5% (56 PwH-Y) of the patients were in the prophylaxis program in the treatment centers; however, only about half (46.2%) of the PwH-Y stated that they received regular infusion for prophylaxis 2 or 3 times in a week and a high percentage (71.4%) of their relatives were in the opinion

Table 2. General Characteristics of Persons With Hemophilia of Generation Y and Their Non-Hemophiliac First-Degree Relatives.

	PwH-Y n = 64	Relatives n = 56
Sex, Male/Female, n/n	62/2	17/39
Age, years; mean (range)	28 (16-42)	47 (25-68)
<18, n (%)	3 (4.7)	–
18-36, n (%)	60 (93.8)	5 (8.9)
>36, n (%)	1 (1.5)	51 (91.1)
Relationship, n (%)		
Father	–	15 (27.8)
Mother	–	35 (62.5)
Spouse	–	3 (5.4)
Brother	–	2 (3.6)
Grandmother	–	1 (1.8)
Disease, n (%)		
Hemophilia A	53 (82.8)*	–
Hemophilia B	9 (14.1)	–
vWD Type 2	1 (1.6) (female)	–
Factor X deficiency	1 (1.6) (female)	–
Type, n (%)		
Severe	56 (87.5)#	–
Moderate/Mild	8 (12.5)	–
Treatment, n (%)		
Prophylaxis	56 (87.5)	
On demand	8 (12.5)##	
Location, n (%)		
Istanbul	28 (43.8)	28 (50)
Antalya	13 (20.3)	17 (30.4)
Tekirdag	5 (7.8)	7 (12.5)
Konya	11 (17.2)	8 (14.3)
Trabzon	7 (10.9)	6 (10.7)

PwH-Y: persons with hemophilia of Generation Y; vWD, von Willebrand's disease.
* 3 patients with inhibitor.
# 50 patients with hemophilia A and 6 patients with hemophilia B.
3 patients with hemophilia A, 3 patients with hemophilia B, 1 patient with vWD and 1 patient with Factor X deficiency.

that hemophiliacs were adherent to treatment. Accordingly, the difference between the rate of adherence to treatment reported by the patients and that perceived by their relatives was significant ($p \leq 0.05$, Figure 1).

Evaluation of the opinions of the groups about difficulties related to adherence to treatment revealed that vascular access problems (60%) ranked first among the reasons for non-adherence to regular prophylaxis, followed by treatment process-emotional difficulties (16.9%) in the PwH-Y, and difficulties concerning accessibility of drugs (15.4%) (Figure 2). These rates were 25%, 8.9%, and 17.9% respectively, in their relatives.

The components that the hemophiliacs and their relatives need most were support for accessibility of drugs and treatment ranked first and followed by emotional support both in the PwH-Y (41.1% and 26.1%, respectively) and the relatives (45% and 32.5%, respectively) (Figure 3).

Evaluation of the social media usage of the hemophiliacs and their relatives revealed that the rates of using Facebook,

Instagram, and YouTube in the PwH-Y were 85.1%, 83.4%, and 74.1%, respectively. These rates were observed to decrease to 55.0%, 32.4% and 21.1%, respectively in the relatives (Figure 4).

Discussion

The people born between 1981 and 1999 are defined as Generation Y.[1,2] According to the 2014 data of WFH, there are 4,860 registered Hemophilia A and 878 Hemophilia B patients in Turkey with the 47% of the hemophilia A patients and 45% of the hemophilia B patients being the members of Generation Y (Table 1).[6] For the success in the management of PwH-Y, which accounts for the substantial proportion of hemophiliac
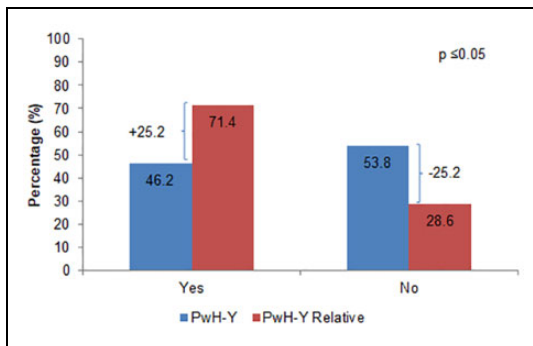

Figure 1. Rates of adherence to regular infusions for prophylaxis reported by the persons with hemophilia of Generation Y (PwH-Y) and perceived by their relatives. P ≤ 0 .05 stands for the difference between the rate of adherence to treatment reported by the patients and that perceived by their relatives.

population, it is important to recognize this generation, to know its characteristics, and to be in agreement and cooperation with this generation. The characteristics of Generation Y include Internet and technology dependence, high ego, continuous and persistent demand, a short attention span, chronic boredom, a tendency for globalization, being open to change, a tendency for self-management, and belief in (perception of) capability of doing everything.[8-10] The members of Generation Y differ significantly from the members of other generations. These differences also likely apply to the PwH-Y as hemophilia is a chronic disease and requires a lifelong management. However, non-adherence to treatment manifests itself as an important problem in the management of hemophiliacs, as does in the other chronic diseases.[11,12]

It is known that the rate of adherence to treatment is nearly 50% among European adolescents with chronic illnesses.[12] In Europe, of the hemophiliacs, 70% modify the prophylaxis regimen on their own and 69% discontinue prophylaxis regimen without notifying their physicians.[13,14] In the present study, although 87.5% (56 PwH-Y) of the patients were in the prophylaxis program in the treatment centers, the rate of adherence to treatment was determined to be 46.2% in the PwH-Y. Nevertheless, adherence to treatment by the PwH-Y was perceived to be higher (71.4%) by their relatives. This is a critical finding indicating that adherence to treatment in chronic diseases is difficult also in the PwH-Y.

The main reasons for non-adherence to treatment among European hemophiliac adolescents are forgetfulness and not being able to spare enough time for treatment.[13] However, in the present study, vascular access problems (60%) as well as the difficulties concerning accessibility of drugs (15.4%) were
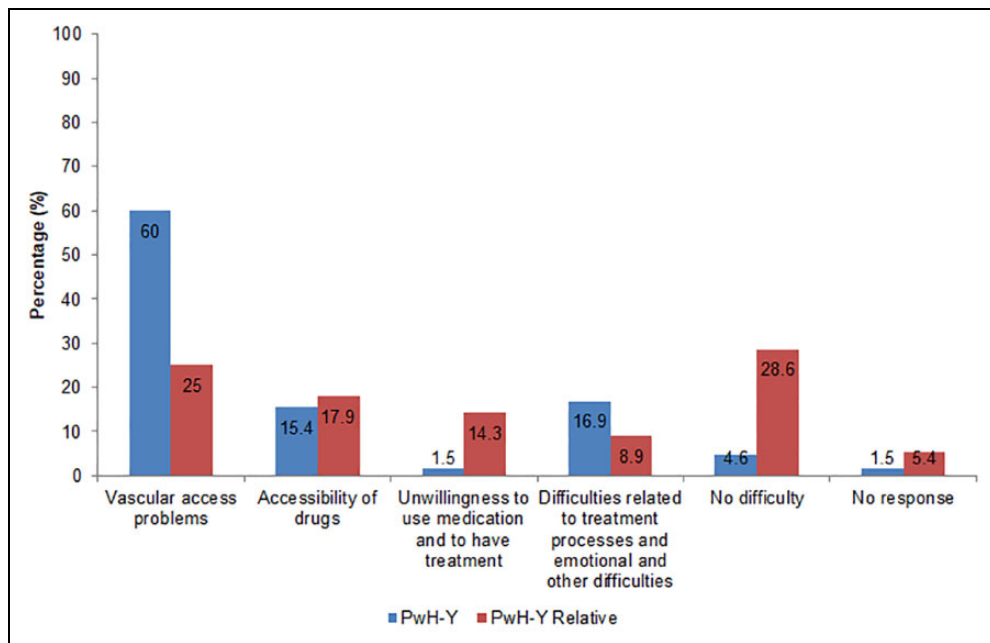

Figure 2. Distribution of the difficulties related to regular treatment according to the persons with hemophilia of Generation Y (PwH-Y) and their relatives.
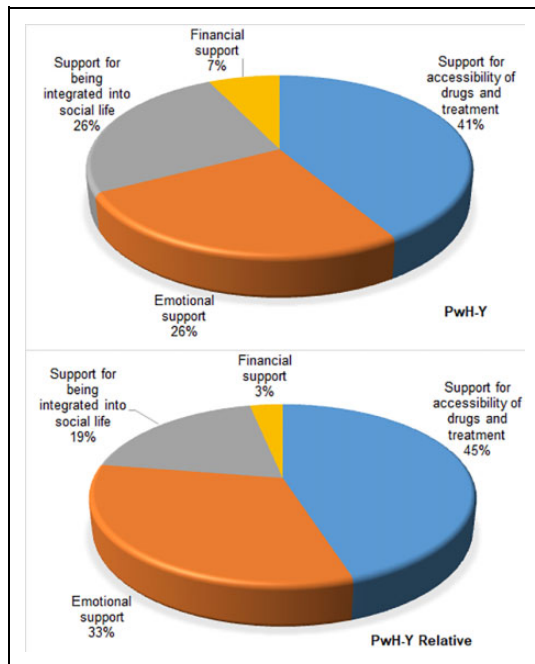
Figure 3. Distribution of the components that the persons with hemophilia of Generation Y (PwH-Y) (A) and their relatives (B) need most.
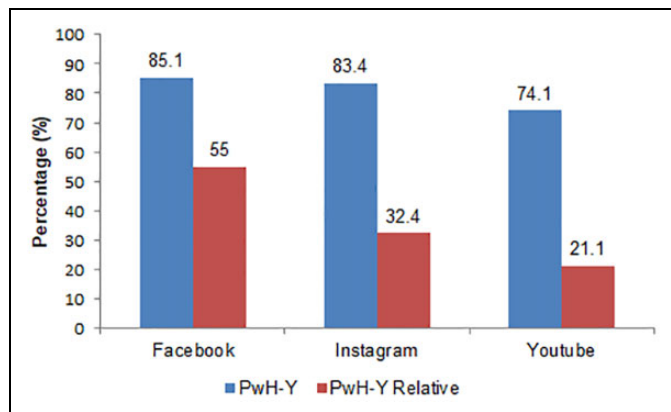

Figure 4. Distribution of the most preferred social media platforms by the persons with hemophilia of Generation Y (PwH-Y) and their relatives.

reported by the patients as the main reasons for non-adherence to treatment.

The hemophiliacs of Generation Y do not pay enough attention to the continuity of prophylaxis, which is the gold standard. Taking the findings of the present study into account, it was concluded that PwH-Y should have a close relationship with their physicians.

In the study, the primary requirement of the PwH-Y and their relatives was found as support for accessibility of treatment (41.1% and 45%, respectively), followed by emotional support (26.1% and 32.5%, respectively), and support for being integrated into social life (25.6% and 19.2%, respectively). These findings suggested that support for accessibility of drugs

and treatment should not be the only goal in disease management but also a comprehensive care service and adequate attention and education are needed. Moreover, it was concluded that the "Personalized Treatment Approach," which takes the living conditions and activities of patients into account and may allow involving patients in treatment, is of importance for treatment planning. By this way, the possibility of achieving the targeted treatment success would be increased.[15] Accurate, comprehensive, and personalized education may provide another significant benefit that it enables the hemophiliacs to spend more time outside home. Hence, these patients could maintain their lives, involve in social activities such as sports and travel, and become more active in life. In their review, Skinner et al.[16] described a new treatment model developed through collaboration between clinicians and patients, which aimed at functional cure through 7 treatment milestones in a stepwise manner and at health equity with a parallel set of patient-reported outcomes. They have suggested that the described treatment model will transform the currently used conservative approach for PwH, which is achieving factor VIII levels of $\geq 1\%$, to a stepwise approach that can fulfill patients' expectations by providing freedom for PwH from lifestyle and medical restrictions caused by hemophilia and by allowing them to choose their level among the proposed treatment milestones.[16]

Communing with technology and the Internet is one of the main characteristics of Generation Y.[17] The hemophiliacs, who live a relatively passive life, take the benefit of social media primarily to have information about everything, to create a social environment, and for entertainment. Thus, the rate of the Internet usage is also high among the PwH-Y. In the present study, the rates of using Facebook, Instagram, and YouTube in the PwH-Y group were determined as 85.1%, 83.4% and 74.1%, respectively. In recent days during which we are all experiencing the coronavirus disease 2019 (COVID-19) pandemic, it was observed during the lock-down period (March-May 2020) in Turkey that social media, which is considered a reliable source of information for the young generation, replaced traditional media, and the programs where specialist physicians discussed the pandemic on television were watched at a high rate and found to be credible. However, it has been then realized that this generation have started to rely on information shared on social media after the lock-down period. Nevertheless, frequent use of the Internet for entertainment, communication, and to obtain information exposes the individuals to misinformation.[18] The hematologists experienced in hemophilia has critical role for accessing correct information through the Internet. Both communication programs and current scientific programs in the Internet provided by hemophilia treatment centers and associations would enable hemophilia patients to access more accurate information. Additionally, based on a recently published article emphasizing the necessity of telemedicine which enables the delivery of remote healthcare to patients especially for those with chronic disorders such as hemophilia and especially for the recent COVID-19 pandemic,[19] hemophilia treatment centers and associations may

consider telemedicine to provide accurate information and healthcare for hemophilia patients.

In conclusion, in order to increase the treatment success in the PwH-Y, the treatment should be personalized and shaped based on the personal requirements. The patients should be involved in decision making process at each step, and conventional room meetings should be established in education platforms where the patients could access correct information. In addition, the number of social media programs in the Internet should be increased and, in particular, the contribution of prophylaxis to the quality of life should be highlighted in all platforms. Preferably, the first step prior to all these actions should be enabling the "Hemophilia Treatment Centers," where a multidisciplinary team that connects closely with hemophiliacs and their relatives and provides care and service to meet any requirements under the leadership of an experienced hematologist on hemophilia, to become widespread.

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