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ORIGINAL RESEARCH

Analysis of the Reasons for Non-Uptake of Predictive Testing for Huntington's Disease in Spain: A Qualitative Study

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Abstract Children of persons affected by Huntington's disease (HD) have a 50% chance of inheriting the disease. Genetic testing in Spain is offered to individuals (presymptomatic test) or mothers of fetuses (prenatal) who run the risk of suffering from HD. The objective of this study is to analyze the factors that influence the decisions of adult children of persons affected with HD regarding predictive testing. A qualitative research methodology was used involving 4 focus groups (FGs) made up of adult children of persons with HD in different cities in Spain. The results of the study showed that over half of the focus group participants were inclined to decline genetic testing. The main explanatory determinants for taking or not taking the predictive test are: Maturity of the individual at risk, which was directly related to age; Ability to cope with a positive test result; Experience of living with HD sufferers; Information about testing and psychological support; Attitude of the family; Social visibility of genetic testing; Personality and temperament of each subject at risk of HD. These results imply that these factors should be analyzed in more detail in quantitative studies in order to help the Spanish Department of Health understand why some children of parents with HD decline genetic testing, so that they may and apply these data when creating specific policy regarding this issue.

Keywords Huntington's disease · Genetic testing · Explanatory factors · Qualitative method

Huntington's disease (HD) is a neurological, degenerative, hereditary-autosomal dominant disease (Timman et al. 2005). It is characterized by movement and psychiatric disorders, mainly affecting mood and cognition. HD generally presents between 30 and 50 years of age, with an average course of 15–20 years. Immediate causes of death are often infections, most frequently pneumonia (DiMaio et al. 1993).

HD remains an incurable disease, although existing pharmaceutical treatments help alleviate some symptoms (immobility, depression, apathy, irritability…). The effectiveness of treatment varies from one patient to another. However, in all cases treatment becomes less effective as the disease progresses (Walker 2007).

The prevalence of HD is between 5 and 10 cases per 100,000 inhabitants in Western countries. The yearly incidence of HD is 1 to 4 cases per million inhabitants (Harper 1992). In Spain, the only study available is for the city of Valencia, which shows a prevalence of 5.38 per 100000 inhabitants (Burguera et al. 1997), which is consistent with data from other European countries (Pringsheim et al. 2012; Hoppit et al. 2011).

HD is inherited in a dominant pattern. Therefore, progeny of a person affected by HD have a 50% chance of inheriting the disease (Tibben 2007), although those people with 36 to 39 CAG repeats may or may not develop the signs and symptoms of HD, while people with 40 or more repeats almost always develop the disorder (Panegyres and Goh 2011).

Although direct gene testing has been used for HD since 1993 (Bernhardt et al. 2009), such testing has recently been regulated by the Spanish Law on Biomedical Research (LBR) (Law 14/2007, issued on July 3, 2007). A predictive test is

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offered to the asymptomatic individual (pre-symptomatic test) or the mother of a fetus (prenatal) whose parent suffers from HD (Tibben 2007). Genetic testing in Spain entails a process that includes: a blood analysis, a neurological exam, a session with a geneticist, a personality test and a psychological interview. The genetic test cannot predict the age of onset, symptoms, severity or rate of progression (Ha and Fung 2012). If a neurological exam shows symptoms of HD, the subject is considered symptomatic, and is conclusively diagnosed as an individual with HD. Conversely, persons who have been diagnosed positive in a predictive genetic test but do not show symptoms will only be considered carriers of the mutation at that point in time (Paneque et al. 2012), i.e., the persons diagnosed positive will certainly develop HD in the future, although for a time they will be able to live symptom-free (Taylor 2005).

Studies related to the uptake or declining of predictive genetic testing by those at risk for HD show varied results; we will immediately proceed to comment on some of these studies. Craufurd et al. (1989) differentiated between individuals at risk for HD who were offered predictive testing and those who spontaneously sought predictive testing; acceptance rates were highest in the second group (more than 50%) than in the first group (15.5%). A study conducted in Canada showed testing uptake between 1987 and 2000 varied from 12.5 to 20.7% depending on the province studied (Creighton et al. 2003); in the Netherlands, another study showed that testing uptake between 1987 and 1997 was 24% (Maat-Kievit et al. 2000). Goizet et al. (2002) conducted a comparative study of presymptomatic testing for HD and autosomal dominant cerebellar ataxias; only 57% of persons who requested genetic testing for HD completed the entire procedure. Two studies from 1987 which specifically analyzed the willingness of at risk individuals to be tested concluded that between 65 and 79% of at risk individuals were disposed to undergo genetic testing (Kessler et al. 1987; Meiseen et al. 1987).

Other studies have analyzed the impact of genetic testing on the insurance, employment, family and social network of people with HD (Bloch et al. 1992; Bombard et al. 2009; Erwin et al. 2010; Goh et al. 2013; Nagaraja et al. 2006; Penziner et al. 2008; Sobel and Cowan 2000; Timman et al. 2004). Main impacts include work-related issues, conflicts with insurance, a loss of membership in the family (separations and divorces), communication problems within the family, reactivation/intensification of old conflicts in the family, and concern over ensuing caregiving.

Further research has focused on coping techniques intended to reduce the effects of the results of genetic testing on the life of the affected person (Richartz-Salzburger et al. 2006; Wahlin et al. 1997). Factors associated with benefits perceived after genetic testing have also been studied (Williams et al. 2010b). Psychological well-being has been compared between persons with early symptomatic HD, gene positive persons, those at risk for HD with unknown gene status, and healthy controls; persons with symptomatic HD have reported greater levels of stress than other groups (Chisholm et al. 2013). Research has also been conducted on the perceptions of genetic testing in offspring of individuals with HD (Binedell et al. 1998a, b; Decruyenaere et al. 1993; Mattsson and Almqvist 1991; Van der Steenstraten et al. 1994). Binedell et al. (1998b) noted the following factors in making decisions regarding testing: a desire for certainty; moral imperatives to clarify one's genetic status (i.e., views of the controllability of the future); family attitudes and norms. Mattsson and Almqvist (1991) and Van der Steenstraten et al. (1994) observed that asymptomatic persons who had passed the mean age of onset within the family were largely receptive to testing, as were those with mild or undetermined symptoms, such as movement disorders. Younger individuals, especially those claiming a good relationship with the affected parent, appeared less interested in being tested. In both studies, all participants demonstrated ambivalence toward genetic testing and an inclination to change opinions over time. Therefore, there were persons who sometimes wanted genetic testing and at other times did not.

In Spain, no studies have been conducted concerning the attitudes toward genetic testing of adult children of individuals diagnosed with HD. Nor have factors related to decisions regarding predictive testing by adult children of persons affected with HD been studied. The study of attitudes toward genetic testing of individuals diagnosed with HD can provide us with an approximation of the reasons why genetic testing is undergone or declined. The lack of previous research, the advisability of lessening the probable impact of results of genetic testing (if the results were positive) on the adult son or daughter of persons with HD and his/her family, and the possibility of decreasing the incidence of people affected by HD have spurred this study on adult attitudes toward genetic testing. Accordingly, the main objective of this study is to investigate the factors that influence adult children of persons affected with HD in their decision regarding predictive genetic testing.

Material and Methods

Our study is cross-sectional and was conducted over a period of 9 months (from January to September 2009), using the qualitative technique of Focus Groups (FGs). We used a qualitative method not to produce generalizable results, but rather to generate insights that can be further tested by more systematic methods (Morse and Field 1995). FGs technique was chosen to facilitate discussion among participants regarding attitudes toward HD and genetic testing, thus enabling researchers to better understand the issues being discussed.

Participants

All participants in the FGs were recruited through volunteer HD associations (informal support groups) in four Spanish cities: Madrid, Barcelona, Burgos and Salamanca. It is common for people affected with HD and their relatives to be connected with such organizations in Spain. The psychologist at each organization was responsible for recruiting adult children of persons affected with HD for this study. The researchers requested of the psychologist at each organization to try to ensure that exactly half of the participants in each FG had taken the predictive test; we tried to adhere to this condition because it was believed that the discussion would be more heterogeneous and enriching (Morgan 1998) and could offer a wider diversity of opinions in favor of and in opposition to the predictive test. Nevertheless, this equal ratio was achieved in only two of the four FGs. The two exceptions were Salamanca FG2, in which 4 participants had not taken the test and 2 had, and in Barcelona FG4, in which 3 participants had taken the test and 4 had not. These differences in the composition of the FGs were caused by the lack of availability of appropriate volunteer participants with these specific characteristics within the associations. Regarding HD onset, we did not gather information about whether the persons who had been diagnosed had begun to show symptoms nor did we determine the level of symptomatology; we considered this information irrelevant to our study. The operative factor was whether they had undergone genetic testing, for this reason we did not take into account the number of CAG repeats in each individual (it was information which we did not have). The final sample consisted of 27 adult children of persons affected with HD, divided into 4 FGs. Burgos FG1 comprised 8 participants, Salamanca FG2 comprised 6, Madrid FG3 comprised 6, and Barcelona comprised FG4 7.

Instrumentation

FGs lasted approximately 1 h and a half and were conducted in an appropriate setting within each city. In Burgos, Madrid and Barcelona, FGs were conducted in cultural centers and in Salamanca in a university building (Social Sciences Faculty), because these spaces were considered neutral, a requirement for each FG (Morgan 1998). At each FG, the facilitator or moderator (the first author of this manuscript) explained the purpose of the meeting, encouraged participation, and assured confidentiality. With the participants' permission, all interviews were audio-recorded and field notes were taken.

Each FG opened with a general question related to opinions regarding the social and medical systems in Spain with regard to HD. These icebreaker questions allowed the participants to settle into the group. The FG questions were not posed in any particular order. The exact wording of the FG questions was not predetermined (although the moderator had a list of themes that would have to be raised and related sample guide questions); rather, the moderator carefully followed the cues of the participants and the discussion that proceeded from those cues. The guiding questions for the FGs were created by the researchers of this study and included the following issues: 1) knowledge of genetic testing in general or specifically pre-symptomatic genetic testing; 2) medical and psychological support; and 3) perception of predictive testing or genetic testing (we will use both terms interchangeably throughout this paper) and explanatory factors.

This flexible FG structure allowed participants to discuss the issues most important to them. When a topic did not arise spontaneously (which rarely occurred), the moderator raised the issue, to ensure that all groups covered the identical list of core topics.

In each FG, all participants participated; if a participant did not contribute, the moderator asked him a direct question (this rarely occurred because the moderator tried to establish to a comfortable atmosphere in which the participants wanted to contribute).

Procedures

The saturation point in each FG occurred when all of the core themes on the facilitator's list had been addressed and the participants were beginning to repeat the contents (Quine 1999).

A qualified audio typist transcribed the recordings of each session, and the researchers checked these transcriptions carefully (Rivera-Navarro et al. 2009). The quotations shown herein are literal statements (cited by Focus Group number and transcript page, e.g., Burgos FG1, 7: Burgos FG number 1, page 7). The direct FG quotes that appear in the text were translated from Spanish to English with alterations made to literal translation to preserve the intended meaning. The procedure of translation of these quotes consisted firstly of a translation from Spanish to English by a native English speaker who is highly familiar with the Spanish language. Subsequently, a bilingual native Spanish speaker translated the text from English back to Spanish, after which another native English speaker translated the text back to English again. This text was then compared to the original text from the FGs, and differences were noted among the three translators. The translators then discussed the discrepancies between the original and the translations, and modifications were made where it was deemed necessary to achieve the most accurate result.

The study protocol was approved by the General San Yagüe Hospital ethics committee. An informed consent form was signed by all participants in the study, and anonymity of the subjects was guaranteed.

Data Analysis

This study was performed using a technique known as Qualitative Description. The goal of qualitative description studies is for the researchers to compile the events that are referred to by the participants in FGs in accessible language. Researchers conducting qualitative descriptive studies adhere to the data and do not inject a psychological interpretation of the language (Sandelowski 2000).

The principle investigator and the two co-authors each systematically read through all of the transcripts and made a list of codes, sub-categories and categories, highlighting all the main categories in the text. Qualitative sequential discourse was the method used for data analysis. This method follows three steps (Hsieh and Shannon 2005): 1) Coding – the words or sentences that express the essence of the discourse are highlighted line by line; 2) Sub-Categorizing – once the coding process is complete, codes are regrouped into new forms, giving rise to conceptual codes or sub-categories, which have a higher level of abstraction than the coding process; 3) Categorizing – categories have a higher level of abstraction than sub-categories, and each category includes several concepts (e.g., in this study, the category "ability to cope" includes concepts such as personality, past life and current way of life). These categories often, although not necessarily always, matched the core topics previously determined by the researchers, i.e., perception of genetic testing, medical and psychological support in genetic testing, etc.

A group of independent raters (two psychologists, two sociologists and one family physician) compared the coding against the transcripts to enhance reliability and the validity of the coding system. Transcriptions and a list of codes, subcategories and categories identified by the authors were provided to the raters, who were requested to match the transcription lines to the codes; the raters could then propose other non-identified codes, sub-categories or categories if they considered it necessary (if they thought a new code, sub-category or category was required to define a transcription). Only one new previously unidentified sub-category was proposed by two raters - *Social visibility as a factor for declining predictive testing* - and the group reached consensus to include this. Although the raters operated independently, they later met on several occasions with the researchers to discuss the codes, subcategories and categories.

Interrater agreement was calculated according to Walker's procedure (Morse and Field 1995). The FG transcriptions were divided into paragraphs that were coded independently by the 5 raters; in every paragraph, the dominant code (the code given by the majority of the raters) was regarded as the correct code. Interrater reliability was considered to be the average of the percentages of reliability of every rater, with an interrater agreement of 90%. The rater with the lowest percentage of agreement was 87.5%, and the sub-category with the lowest percentage of agreement was 86.2%. These were accepted as high percentages, illustrating that the coding, sub-categorizing and categorizing were appropriately consistent.

The data were examined by comparing the results of the different informant groups. Disagreement among participants was taken into account; e.g., there were adult children of persons with HD who had undergone genetic testing who did not consider maturity an important factor in deciding on genetic testing whereas other adult children who had also had genetic testing argued against this opinion because of concerns regarding psychological consequences. These discrepancies were identified in the analysis, expressing the different positions on a specific topic. The researchers believe such differences to be completely normal and likely to be expressed in any single FG.

Results

Our sample comprised 27 adult children of persons with HD (all native Spanish citizens) with a mean age of 37.9 (SD=6.7, range: 24–53). Most (62.9%, n=17) were married, and 55.5% (n=15) were women. Approximately half of the participants (n=14) were college graduates or higher, and the other half were high school graduates or less (n=13). Approximately 63% were currently actively employed (n = 17), approximately 19% were unemployed (n=5), 4 more were students, and the remaining 1 was a stay-at-home spouse. The majority of the participants were members of volunteer HD-related organizations (n=22).

Four different themes emerged across the FGs. Each theme is described in detail below.

Awareness of Genetic Testing

All participants in FGs were familiar with predictive genetic testing and the testing's main characteristics.

In Favor of Genetic Testing

Of all the participants, 48.1% (n=13) specifically expressed that they would have genetic testing and thought that in general, undergoing the testing was beneficial to the adult children of persons with HD and to their relationship with their environment (family, friends and work setting). In this case, the participants believed that being certain about their mutation status could improve relationships with spouses, children and/or colleagues. If the test was positive, the individual could adopt coping strategies; and if the test was negative, the individual could live without that concern. Nevertheless, there were several factors that were noted by these participants as

decisive indicators for wanting or not wanting to be tested. These will be discussed later.

"I think that taking the test might help avoid misunderstandings with my colleagues at work because they may understand what would be happening to me, if I do have the disease" (Madrid, FG3, 4).
"I think predictive testing is a good thing; if you do the test and it comes out positive, your friends or relatives can pick up on the first symptoms and that way they'll help you from the start and it will all be easier" (Madrid FG3, 5).
"I had a boyfriend, and my decision, in my case not for me, but for my boyfriend was to take the test. I don't want to put my boyfriend through something that could happen" (Barcelona, FG4, 6).

Against Genetic Testing

Some participants in FGs thought taking the genetic test would have a direct negative effect on the life of the person who was tested. These participants were apprehensive that a positive result would provoke constant worrying about when the onset of symptoms would occur or that they would attribute any unexpected movement or subjective experience to HD symptoms, however unrelated. Those who had no wish to take the test viewed genetic testing as denying themselves the opportunity to enjoy life, and they did not feel prepared to cope with a positive result.

"The burden would be unbearable, and I still have a long life ahead of me; I think that I would grow very bitter if the test came back positive" (Salamanca, FG2, 3).
"I think you have to live your life, live without thinking about whether or not you have the disease" (Salamanca FG2, 9).
"I consider that taking the predictive test might ruin my life; knowing I am going to suffer a disease could traumatize me so much I wouldn't want to go on" (Madrid, FG3, 4).

Another concern about proceeding with predictive testing was the perceived futility; according to some participants, the impossibility of recovery for people afflicted with HD caused a negative view toward testing. Moreover, the perception of the inability of the Spanish public health system to provide high quality treatment and therapy to people with HD exacerbated these participants' feelings that genetic testing is useless.

"Because the health system doesn't offer any support services, people don't want to take the test. They think, what's the point if it's not going to make a difference? Seeing as, for the moment, there's no cure, they don't see how it would help them" (Madrid, FG3, 13).
"The health system doesn't offer services of any kind to support the recently diagnosed person" (Burgos, FG1, 13).

The final argument against the predictive test was the perception of a lack of psychological support or pre- or post-test counseling.

"Once you've taken the test, if they tell you you have the gene, even if you're not showing any symptoms of Huntington's disease yet, there's this sense of utter emptiness, even if you supposedly have psychological support" (Barcelona, 4FG4, 13).
"Here, in the hospital in Burgos, genetic testing has been conducted without so much as a pre-test psychological assessment, never mind counseling; and what's more, up until recently, test results have been given out over the phone" (Burgos, FG1, 14).

Factors Influencing the Decision to Take or not to Take the Genetic Test

In FGs, participants spoke spontaneously about factors that could determine whether they would undergo genetic testing. These were as follows:

– Maturity, which was directly related to age. In several FGs, when some of the participants spoke about a relative who had taken the test, if the relative was young, he/she was criticized by other participants and the decision was criticized:

"I don't think that an 18-year-old is ready to undergo HD testing yet. I believe that taking the test so young could be harmful, and they still have so much time, there's no need to rush to find out whether they have the gene or not" (Burgos FG1, 17).

– Ability to cope with having the genetic testing. This depended on the individual life experiences of each of the affected persons and their current way of life:

"The ability to grasp the results of the predictive test depends on many things: on your way of life, on your experiences…For example, if you have a chaotic lifestyle and you're prone to depression, you're not going to take a positive result well" (Salamanca FG2, 7).

– Experience with living with individuals who are affected with HD and having accepted this illness (these two

separate domains were connected by some participants in FGs). The years of living with a symptomatic loved one may provide a comprehensive view of the disease and help an individual embrace HD as a "normal life event". This was perceived as a factor that favors having genetic testing. Nevertheless, some participants held the opposite view; they suggested that a lack of experience with sharing daily life with a person with HD may encourage at-risk individuals to have genetic testing because this lack of firsthand experience may have resulted in the absence of a dramatic perception of HD. In this case, not having in-depth knowledge of the evolution of HD could be an influential factor in support of having genetic testing.

"I think that *not* having lived with my father when he began to show Huntington's symptoms gave me the impetus to get tested" (Burgos FG1, 9).
"I think the opposite. I think living with someone with Huntington's and understanding the disease can spur you to take the predictive test" (Burgos FG1, 9).

– Information that explained the advantages of testing and good psychological support to minimize the effect of testing.

"I think information is very important, in the sense of being aware of the chances of suffering from Huntington's and knowing the risk involved with regard to having kids too and how you can pass it on to your child" (Madrid FG3, 7).
"We went to a neurologist to find out what was wrong with my mother. Once she was diagnosed, the whole family was informed of the characteristics of Huntington's disease. That was very important because it meant we understood what my mother had and we saw the disease in another light; we weren't so afraid" (Barcelona FG4, 4).

– Attitude of the family. In some FGs, the feelings of guilt of members of different families were attributed to the hereditary nature of the disease. Thus, members of some families thought having genetic testing would be ill-advised because it increased the feeling of guilt of persons previously diagnosed with HD and indeed of those members of the family who tested negative beside their positively diagnosed siblings.

"I acknowledge my father's bravery, even though he feels very guilty; he feels terrible" (Salamanca FG2, 9).
"My father never wanted to accept it; he couldn't bear to know one of us could have the condition due to hereditary causes" (Burgos FG1, 20).

– Social visibility. Some FGs participants showed reluctance to have genetic testing to avoid having the people in their social environment infer that other members of the family may have HD because of its hereditary nature. This phenomenon alludes to the stigma of HD in Spanish society and was commented on in all FGs.

"There are people who hide it to protect the privacy of the rest of the family; there are even people who admit it: 'I want to get tested, but I don't want anybody to know'" (Burgos, FG1, 9).
"I think there are ethical issues to be taken into account. For example, how do you safeguard the freedom of choice of a sibling who does not want to have genetic testing or ensure that word does not get out in the family that the father or the mother has Huntington's if another sibling, son or daughter wants to be tested?" (Barcelona, FG2, 22).

– Personality and temperament. Some participants of FGs emphasized the unique personality of each son or daughter of a person with HD to explain the different reactions to genetic testing.

"I find it very difficult to judge what one should or shouldn't do. It is a very personal decision that depends on how ready one feels to face the test. There is no recipe for that" (Barcelona, FG4, 6).

Discussion

This paper provides the results of a study based on qualitative methodology that analyzes the attitudes toward genetic testing of adult children of persons affected with HD. All participants were sourced through HD support groups. The study was conducted in four Spanish locations – two large and two medium-sized cities.

Just under half of all participants in FGs (n=13) favored genetic testing and perceived positive repercussions from undergoing this process. The participants in FGs who favored genetic testing spoke only of perceptions, opinions or points of view regarding genetic testing; these attitudes did not involve actual testing uptake. Our study did not explicitly identify whether the opinion in favor of "undergoing genetic testing if it were available" was more supported by tested or non-tested participants. Nevertheless, it is reasonable to believe the tested individuals are more inclined to defend genetic testing. Van der Steenstraten et al. (1994), in a study in the Netherlands in which participants in a predictive testing program were compared with non-participants at

risk, concluded that the non-participants showed a strong negative attitude towards genetic testing.

Just over half of the participants in our FGs perceived genetic testing negatively. They believed testing could cause an obsession with HD or HD-related disorders and reduce perceived life fulfillment and satisfaction. This feeling has been noted in many studies (Wolff and Walter 1992; Van der Steenstraten et al. 1994; Taylor 2005) and indicates a strong cross-cultural tendency regarding PT perceptions in different countries.

Furthermore, the lack of a cure for HD and/or the fear of an unfavorable result of genetic testing also encourages the negative perception of genetic testing, consistent with previous studies (Babul et al. 1993; Creighton et al. 2003; Decruyenaere et al. 1995; Taylor 2005; Van der Steenstraten et al. 1994). The other main argument used by participants in this study to rationalize their mistrust of genetic testing was a lack of confidence in the Spanish public health system and the pre- and post-test psychological support provided. The Spanish public health system is based on curative medicine, mainly in hospitals; there is little encouragement to avail of psychological support services and other multidisciplinary approaches to treat chronic diseases are not encouraged. Although we did not identify any studies that address the mistrust of the health system, several studies did analyze the importance of pre- and post-test psychological support and counselling for HD (Lickleder et al. 2008; Taylor and Myers 1997; Decruyenaere et al. 2003; Huggins et al. 1992). Our data indicate that the perception of a lack of psychological support could create a negative perception and possibly a future declining to participate in genetic testing.

Participants in FGs highlighted several factors as determinants in individuals' decision to have the test. The first factor that we noted was maturity, which was directly associated with age. In some studies, age has been considered a factor in declining genetic testing (Duncan et al. 2008; Taylor 2005; Richards 2006); younger individuals believe they have time to undergo genetic testing, and at present they perceive more disadvantages than advantages to having genetic testing. The present study further confirms this consideration.

The second factor that we showed was the ability to cope with the results of genetic testing. This ability depended on individuals' personalities, their life experiences to date and current way of life. These results have been corroborated in other publications; thus, coping ability has been recognized as a key factor in discouraging participation in HD testing (Decruyenaere et al. 1995; Evers-Kiebooms et al. 2000; Tibben et al. 1993). Moreover, the most common profile of the individuals in our study who have undergone testing was married women (or women in a stable relationship) who already have children, which is consistent with previous studies from Creighton et al. (2003) and Decruyenaere et al. (1995). These women have apparent stability in their lives.

The third factor was whether the person had had the experience of living with a loved one with HD. In our FGs, some participants thought living with an individual with HD could encourage genetic testing whereas other participants were of the opinion that the close everyday contact with persons affected by HD could discourage participation in genetic testing. Nevertheless, the nature of each family's experience with the development of the disease is relevant to predictive test decision-making (Chapman 2002; Cox 1999; Cox and McKelling 1999).

The fourth factor is the information received regarding genetic testing: advantages, effect on daily life, coping strategies, etc. Other studies have shown that a lack of information regarding genetic testing is a discouraging factor (Chen et al. 2012; Duncan et al. 2008). A recent study conducted by Sizer et al. (2012) in South Africa indicates that a lack of awareness of genetic testing is a cause of not having genetic testing, although in European countries, according to several studies, the adult children of persons with HD are generally aware of the existence of genetic testing (Lickleder et al. 2008; Sarangi et al. 2005).

The fifth factor raised is the attitude of the family, specifically the feeling of guilt the hereditary nature of HD engenders in some members of the family. Although we have identified only one previous study that mentions feelings of guilt in HD (Codori and Brandt 1994), this feeling has also been observed with regard to other hereditary diseases, for example, Tourette's syndrome (Rivera-Navarro et al. 2014).

The sixth factor is the social visibility that genetic testing implies, which may be a serious impediment to genetic testing in a close, family-based society such as Spain's (Jurado and Naldini 1996). This phenomenon of keeping HD a secret (and the implications of genetic testing on this) can cause conflicts and rifts in the family and as in the case of the previous factor, also alludes to the stigma of HD (Loi and Chiu 2012; Morrison 2010) and the possibility of discrimination against individuals on the basis of test information. This last argument has been discussed by several authors (Alper et al. 2002; Bombard et al. 2009; Erwin et al. 2010; Goh et al. 2013; Otlowski et al. 2002; Penziner et al. 2008). However, in our FGs, the discrimination is related chiefly to the stigma within the family, referring to the specific characteristics of Spanish families. In Spain, no previous studies have analyzed the perception of predictive testing for HD although published studies regarding other disorders assert that related social problems may be different in other countries because of cultural differences (Del Pino-Casado et al. 2011). In the Spanish context, just 40 years ago the family was considered a collective project, the extended family was barely distinguished from the nucleus. In recent years there has been a considerable rise in individualism and the consolidation of individual identities and there is now social freedom in the way of conceiving and organizing life in a couple and within one's own family.

However, the traditional Spanish family concept lives on in deep-rooted attitudes and opinions; thence social control exerted between different family members (a remaining characteristic of the traditional extended family) is still present in Spain (Meil 2000). Close relationships among various family members such as grandparents, aunts, uncles, cousins, and nephews remain important (Cooke 2009; Maya Jariego 2009). This profile of the Spanish family implies that social control within the family may be more intense than in other countries (Ros and Schwartz 1995), which may be reflected in issues such as predictive testing for HD. Stigma and genetic discrimination occur across many cultures and countries in which prevalence among immediate family members is common (Williams et al. 2010a); nevertheless, in countries in which relationships among different family members are quite close, stigma is also prevalent in the extended family (Losada et al. 2003; Rivera-Navarro et al. 2014). This aspect of HD has not been highlighted in the literature and should be considered with regard to any psychological work undertaken with the families of persons with HD. Nevertheless, the influence of cultural characteristics on genetic testing has been described in several previous studies (Chen et al. 2012; Sizer et al. 2012), and these studies have noted that the cultural environment of each place could determine attitudes toward genetic testing.

Finally, personality and temperament are noted as important factors in FGs to be considered in genetic testing. In another recent study, this type of factor has been shown to influence the suitability of genetic testing candidates (Uhrová et al. 2013).

Some factors, which have been studied in depth elsewhere, were not analyzed in our FGs, such as distress; distress and its causes (the perceived proximity of the disease onset and its interaction with risk perception) (Decruyenaere et al. 1993) is known to influence the decision to undergo genetic testing. A "new" concept called "future discounting of genetic testing" was not examined in our study either; this concept could be considered a factor. Future discounting of genetic testing refers to the low utilization of genetic testing because of the ensuing thoughts of a man or woman with an HD-affected parent: "The risk of my child being affected with HD is only 1 in 4, and even if the HD gene is inherited by my child, the disease may not appear for another 50 years, and during that time medical science could come up with a cure" (Shulman and Stern 2014).

Finally, ambivalence toward genetic testing was also common among participants in FGs. In many cases, the same individual was not clearly against or in favor of genetic testing; he or she would sometimes speak favorably of genetic testing and at other times criticize genetic testing. These data are consistent with other studies (Decruyenaere et al. 2007; Mattsson and Almqvist 1991; Van der Steenstraten et al. 1994).

Regarding the limitations of the study, we note the following: 1) All participants in FGs belonged to or were in contact with HD support groups; members of support groups have access to more information regarding the disease and predictive testing than non-members and were therefore more likely to show a greater interest in genetic testing than at–risk persons who do not belong to such organizations and lack the definitive support of other members (Binedell et al. 1998a, b). This suggests that the opinions and perceptions of the FGs' participants may be biased. 2) We acknowledge that we have not mentioned theoretical approaches to genetic testing such as different models of decision-making applied to predictive testing in HD (social cognition models of decision-making or stage model frameworks). To exclude these approaches was a conscious choice based on the sociological orientation of our study; we have not considered it opportune to directly analyze perceptions of genetic testing for HD. 3) Finally, we must note that data were collected 6 years ago; since that time, some changes may have occurred regarding genetic testing. Nevertheless, we have no evidence of such potential changes; for instance, Sarasola and García (2014), in a recent Spanish book related to genetic advice on several neurologic diseases, consider problems similar to those present 5 or 6 years ago. Furthermore, legislation has not been modified, and the public health system in Spain has not increased information regarding genetic testing for HD because of the country's economic problems. We can thus assume that the situation with regard to genetic testing in HD has scarcely changed. Keeping these limitations in mind, we must consider this qualitative study as a first step to studying this topic in the future; in a follow-up qualitative study, more FGs may be included, some FGs comprising only persons who have had genetic testing and some FGs comprising only persons who have not had genetic testing. Other FGs could be comprised of children with positive compared with negative test results. In this next study, more cities may also be included, which would produce a deeper analysis of the nationwide perception of genetic testing. Finally, quantitative studies are required to delve into aspects only touched upon in this study such as the social stigma of HD, and to study a representative sample in order to generalize results.

Despite these limitations, of which we are aware, we consider this study to have some pertinent strengths: 1) FGs are possibly the best qualitative technique to define the perception of genetic testing because they allow for the inclusion of natural paradox and contradictions. Moreover, research has shown that at-risk individuals explain their attitudes quite differently when asked to spontaneously give their reasons for wanting or not

wanting testing as opposed to responding to a multiple choice questionnaire (Wolff and Walter 1992). 2) That all participants of our study were associated with HD support groups could mean that they were aware of all genetic testing issues; this statement is based on a publication that affirms that participation in support groups enables members to have more complete knowledge of issues related to HD (Coulson et al. 2007); therefore, these participants have sufficient information to speak knowledgeably about this topic. 3) In our study, specific determinants of perceptions of HD predictive testing are identified; some of these factors, such as a lack of confidence in the health system or fear of the social visibility of HD, have not been mentioned in other research.

Conclusions

Our findings are not surprising with regard to previously published data in this area. Nevertheless, our findings are of interest and importance because they suggest that future investigations should verify whether in Spain there may be a less of a tendency to request genetic testing for HD than in other countries. The main reasons for this suggestion are the presence in our study of a negative perception towards the public health system, including distrust in genetic-testing psychological support. These factors should be analyzed in more detail using quantitative studies, because these issues may be hindering the early detection of HD. These factors, if they were confirmed in a quantitative study, could be adding to the increasing discrediting of the Public Health System because of economic crises in Spain (Sermeus 2012). We must remember that in Spain, genetic testing for HD is conducted by the Public Health System, and despite Spain's complying with current European Union regulations regarding genetic testing (Ruíz 2004) and the obligatory confidentiality of the results (Ramos-Arroyo 1998), the Spanish population is inclined to distrust this type of socially sensitive test (Ruíz 2004).

In addition, further investigations must be made in accordance with other results shown to confirm some interesting data our study describes, such as the feeling of guilt of a parent affected by HD regarding his/her children (caused by their being carriers of HD) and the influence of stigma in declining predictive testing.

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Animal Studies No animal studies were carried out by the authors for this article

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