Research Participation: The View of Persons At Risk and Persons with Premanifest Huntington's Disease

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Introduction

There has been great progress in Huntington's disease (HD) research over the last years [1]. Yet, effective disease-modifying therapies that can change or halt HD before the onset of any disabling symptoms are still unavailable [2]. Scientific breakthroughs in this field require an active, informed, and lasting commitment from HD families with research. However, they are traditionally less involved and heard in studies and trials [3]. Specifically, those at risk for HD whose genetic status is unknown (HDRisk) and those that tested positive for HD and are in a so-called premanifest or prodromal disease stage (PreHD) should be engaged in this process.

Our aim was to determine which factors affect the willingness of persons with HDrisk and PreHD to participate in research and check for any differences on this topic related to HD status.

Methods

The European Huntington Association (EHA) created an anonymous online survey to collect information about the perceptions and experiences of research participation among persons with HDRisk and PreHD across Europe (Figure 1). The survey was created through the SurveyMonkey platform [4], contained twelve questions and took around eight minutes to complete. The survey was translated and made available in 12 languages: Dutch (with variants for use in Belgium and The Netherlands), English, French, German, Italian, Norwegian, Polish, Portuguese, Russian, Slovakian, and Spanish. Persons with HDRisk and PreHD were compared on their answers to questions about research experience and knowledge, sources of information about research, factors preventing and facilitating study participation, and the importance of reasons for getting involved and not getting involved in studies and trials.

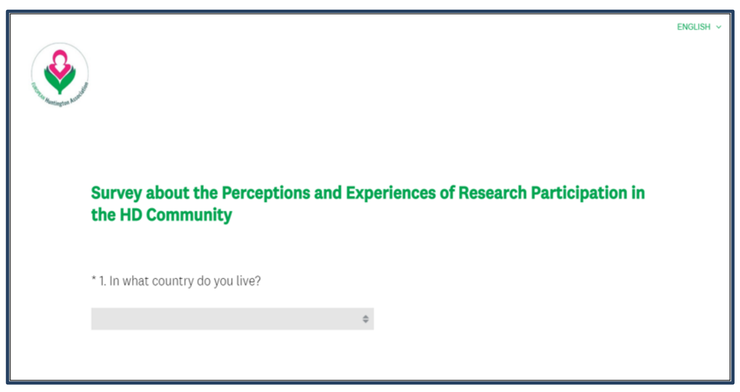

Figure 1 EHA Online Survey

Results

The survey was filled by 263 persons with HDRisk and 262 persons with PreHD, from 27 countries (Table 1).

While the overall motivation of both groups to engage in research was high, respondents with PreHD reported significantly more research experience and better research knowledge than respondents with HDRisk (Table 2). Patient organizations were consistently identified by the two groups as trusted sources for information about studies and trials (Table 2). The motivation to participate in research was influenced by both personal factors, such as psychological, familial, or financial stability, and interpersonal factors, such as access and support from health professionals (Figure 2). Notably, the willingness to participate in research differed according to HD status: respondents with HDRisk identified more barriers to research participation and the need for more support to engage in studies compared to respondents with PreHD (Figures 2 and 3). Additionally, and in accordance with the latter result, respondents with PreHD gave more importance to the reasons for getting involved in research and respondents with HDRisk gave more importance to the reasons for not getting involved in research (Figure 3).

Demographics				
	Total n= 525	***HDRisk n= 263***	***PreHD n= 262***	***Chi-Square HDRisk vs PreHD***
	%	*%*	*%*	*χ2 (p-value)*
Gender				
Female	71.6	73.4	69.8	0.808 (0.369)
Male	28.4	26.6	30.2	
Age Interval (years)				
18 to 24	8.6	12.5	4.6	10.631 (0.001) **
25 to 34	29.5	27.8	31.3	0.791 (0.374)
35 to 44	33.1	35	31.3	0.804 (0.370)
45 to 54	18.3	18.3	18.3	0.000 (0.984)
55 to 64	8.4	5.7	11.1	4.920 (0.027) *
65 to 74	1.5	0.8	2.3	2.046 (0.153)
75 or older	0.6	0	1.1	3.029 (0.082)
Education Level				
Did Not Attend School	0	0	0	-
Completed Primary Education	1	0.4	1.5	1.829 (0.176)
Completed Secondary Education	20.4	18.3	22.5	1.473 (0.225)
Graduated from High School	28.4	27.8	29	0.101 (0.751)
Graduated from College	26.5	29.7	23.3	2.740 (0.098)
Completed Graduate School	23.8	24	23.7	0.006 (0.938)

Table 1 - Demographic Characteristics
* statistically significant $p \leq 0.05$ ** statistically significant $p \leq 0.01$

Experience, Knowledge and Information about HD Research				
	Total n= 525	***HDRisk n= 263***	***PreHD n= 262***	***Chi-Square HDRisk vs PreHD***
	%	*%*	*%*	*χ2 (p-value)*
Previous HD Research Experience				
Yes	31.2	16.3	46.2	54.384 (< 0.001) **
No	68.8	83.7	53.8	
Knowledge about HD Research				
Not Good	5.3	6.5	4.2	1.334 (0.248)
Should be Better	33.3	39.2	27.5	8.061 (0.005) **
Satisfactory	24.6	26.2	22.9	0.788 (0.375)
Good	28.6	23.2	34	7.467 (0.006) **
Excellent	7.8	4.9	10.7	6.015 (0.014) *
Do not want to know about HD research	0.4	0	0.8	2.015 (0.156)
Sources of Information about HD Research				
Internet	78.5	78.7	78.2	0.017 (0.897)
Television	4	2.7	5.3	2.458 (0.117)
Press/Newsletters/Flyers/Booklets	19.8	22.4	17.2	2.284 (0.131)
HD Associations and/or Support Groups	52	51	53.1	0.233 (0.630)
Health Care Professionals	25.5	23.2	27.9	1.505 (0.220)
Family Members	25.3	30.8	19.8	8.321 (0.004) **
Not interested in HD research information	0.4	0.4	0.4	0.000 (0.998)

Table 2 – Experience, Level of Knowledge and Sources of Information about HD Research
* statistically significant $p \leq 0.05$ ** statistically significant $p \leq 0.01$

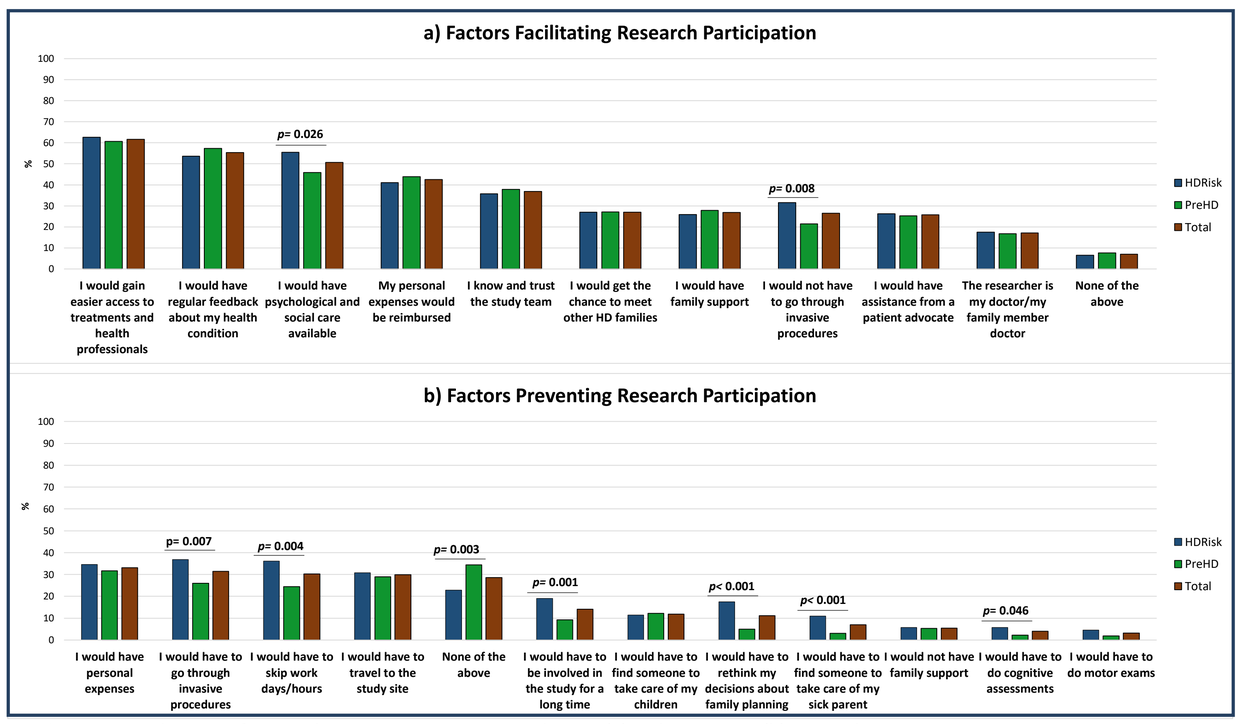

Figure 2 – Moderators of Research Participation: Factors Facilitating (a) and Preventing (b) Research Participation (percentage of participants that selected each factor)

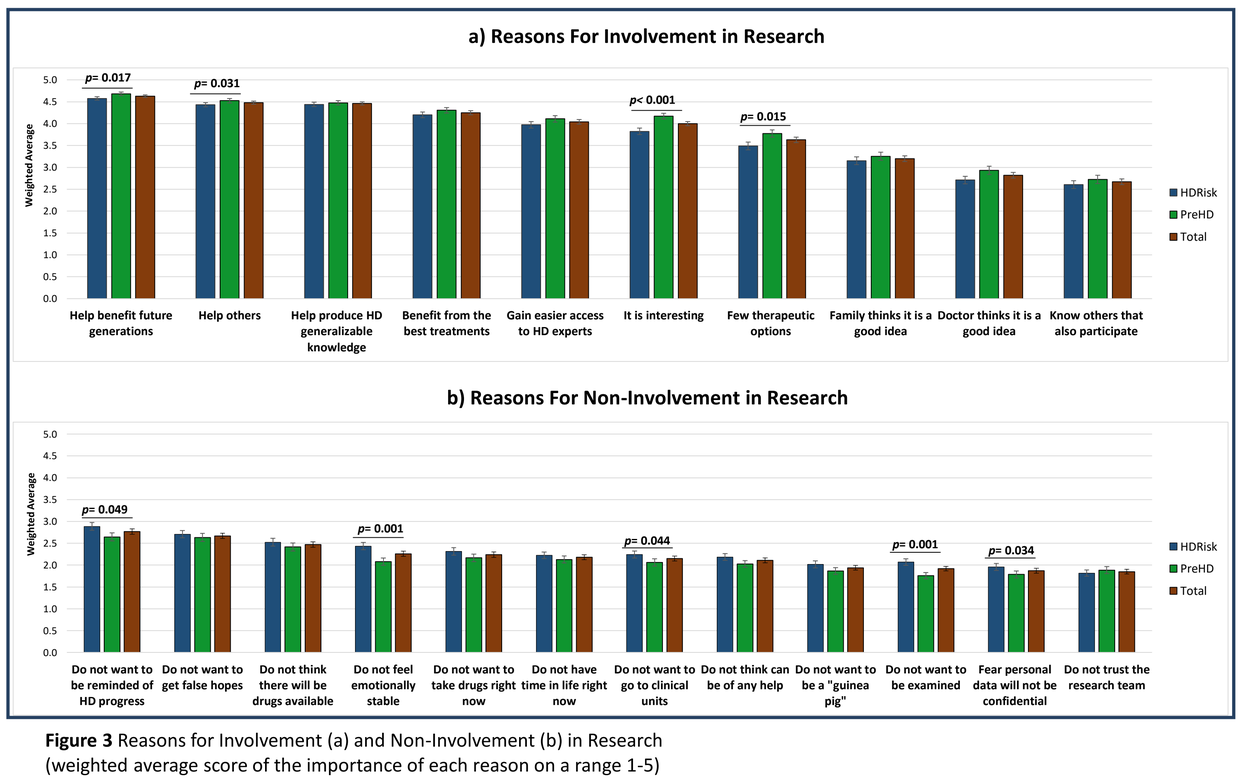

Figure 3 Reasons for Involvement (a) and Non-Involvement (b) in Research (weighted average score of the importance of each reason on a range 1-5)

Key Findings and Conclusions

- Motivation to take part in studies is high, despite limited research experience and literacy
- This motivation is strongly influenced by:

Time/Financial Constraints · Invasive Procedures · Professional and Familial Support · and, importantly, by HD Status

- Solid knowledge base for country-specific and group-specific action planning within Moving Forward
- Patient organizations have a key role in fostering research engagement through education and support
- Tailored interventions seem crucial to promote a meaningful participation of specific HD groups in research

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