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Title page

•Succinct title:

Presymptomatic genetic testing for hereditary cancer in young adults: a survey of young adults and parents

Running head: Genetic testing for young adults

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Abstract

Presymptomatic testing for hereditary cancer syndromes should involve a considered choice. This may be particularly challenging when testing is undertaken in early adulthood. With the aim of exploring the psychosocial implications of presymptomatic testing for hereditary cancer in young adults and their parents, a cross-sectional survey was designed. Two questionnaires were developed (one for young adults who had considered presymptomatic testing, one for parents). Questionnaires were completed by 152 (65.2%) young adults and 42 (73.7%) parents. Data were analysed using descriptive statistics, inferential testing, and exploratory factor analysis and linear regression analysis. Young adults were told about their potential genetic risk at a mean age of 20 years; in most cases, information was given by a parent, often in an unplanned conversation. Although testing requests were usually made by young adults, the majority of parents felt they had control over the young adult's decision and all felt their children should be tested. Results suggest that some young adults did not understand the implications of the genetic test but complied with parental pressure. Counselling approaches for presymptomatic testing may require modification both for young adults and their parents. Those offering testing need to be aware of the complex pressures that young adults can experience, which can influence their autonomous choices. It is therefore important to emphasise to both parents and young adults that, although testing can bring benefits in terms of surveillance and prevention, young adults have a choice.

Key Words.

Decision-making, genetic counselling, hereditary cancer, young adults, parents, presymptomatic genetic testing, familial cancer syndromes

INTRODUCTION

Presymptomatic genetic testing (PST) involves testing to determine if a person has inherited a gene variant that causes a familial condition, before the person has any signs or symptoms of the condition[1]. This type of testing is available for a number of heritable genetic disorders, including some hereditary cancer syndromes[1]. The results of PST for hereditary cancer syndromes may allow individuals to engage in healthy lifestyle choices or seek early treatment for symptoms[2,3]. Some key challenges associated with the transition from adolescence to adulthood can include completing education, beginning full-time employment, forming romantic/sexual relationships, marriage and becoming a parent: the impact of testing may affect, and be affected by, each of these events.

A variety of psychosocial responses have been observed in people who have chosen to be tested[4–6]. Various guidelines and position papers have been produced on PST in minors[7]. It is clearly suggested that undergoing PST too early in life may increase the risk of unfavourable impact, and, therefore, the appropriate age to undergo PST is still a matter of debate[7–9]. For these reasons, PST for adult-onset disorders is not generally recommended for those aged less than 18 years, unless it is in a child's best interests either in terms of immediate relevance for their health or because it involves psychological or social benefits[10]. Although the definition of a young adult (YA) can be extremely broad and is not often clear in terms of one specific age group, the definition proposed by Rindfuss[11] (18- 30 years) was used: 18 years is an age that is often recognized in law and 30 years often represents time for taking stock in life. Prior to testing, YA need to be aware of the potential risk to them of

hereditary cancer, and this is usually disclosed by their parents[12–14]. A systematic review[15] on this topic indicated that many YA or adolescents (14- 30 years) grew up with little or no information concerning their genetic risk and that parents had exerted pressure during the testing decision-making process. An empirical qualitative study[16] conducted in Italy indicated that YA made a decision to be tested before approaching genetic services, and had not realised that they could use genetic counselling to make a choice. However, the process of genetic counselling enabled them to act more autonomously and to adapt to the results. This study was designed to build on those results and further explore the psychosocial implications of PST for hereditary cancer in YA and their parents. Specific objectives were to investigate how YA interpret PST, the reasons for the YA's decision to undergo testing, the experience of the counselling process of both YA and parents and the influence that parents have both on the choice to be tested and on the YA's decisions after receiving a positive test result.

MATERIALS AND METHODS

The study design was a cross-sectional self-completion survey[17]. This study received ethics approval from both St. Orsola-Malpighi Hospital Ethical Board (198/2015/O/Oss), and Plymouth University Faculty Research Ethics Committee (15/16-519).

Recruitment and participants

To maximise accessibility to the survey, online and traditional methods of recruitment and data collection were used. Although online surveys are a convenient way of collecting data from a wide range of people[18], there is evidence that many members of the Italian population do not use the

Internet regularly[19]. Data were collected using: i) online questionnaires (Italian and English version) uploaded to the Survey Monkey® website (for example using social networks) and ii) paper versions (Italian) of the same questionnaire. Traditional recruitment was used only at St. Orsola-Malpighi Hospital, Italy, and specific ethical approval was obtained for that. Every YA or parent of a YA who had been tested who met the inclusion criteria was invited to take part in the study. Parents and YA who responded were not necessarily related to each other. The surveys were open to respondents between December 2015 and June 2016. Inclusion and exclusion criteria and the recruitment flow-chart are presented respectively in Figure1 and Figure 2.

Questionnaire

Since it was important to investigate both consultands' (YA aged 18-30 years[11]) and parents' points of view, two questionnaires were designed. The questions were based on the results of a systematic review[15] and a qualitative study of YA's experiences of PST[16] and on other similar surveys[20,21]. The questionnaires were written both in Italian and English (English version in supplementary files).

Data analysis

In this cross-sectional study, data were entered into a dedicated database (Statistical Package for the Social Sciences (SPSS) Ver. 21.0 for Windows) (IBM Corporation, Armonk, NY, USA), arranged by variables and analysed using descriptive and inferential statistical tests. Descriptive statistics were used to determine the mean, standard deviation, percentage and frequency of variables. The chi-squared test for independence was used to discover if there was a relationship between

two categorical variables[22]. The Fisher's exact test, the independent t-test and ANOVA were used to analyse the data inferentially[24]. Post-hoc tests were also performed where appropriate[22]. Furthermore, exploratory factor analysis (EFA) was carried out to reduce the number of variables, by identifying a limited number of underlying factors explaining multiple observed variables[23-25]. EFA led to identify 20 factors under 6 main questions (Table 1). The factors were also analysed using descriptive statistics and hypothesis testing to analyse the data inferentially. Simple and multiple linear regression analysis were then used to identify effects of independent variables (dummy-coded) on the factors identified by EFA (dependent variables); detailed data from multiple linear regression are reported in Table 2 (a-e). Throughout the study, results were considered statistically significant when the p-value was less than .05. For further information on the methods used for statistical analyses, see Supplementary files.

Rigour

To ensure rigour, a pilot of the survey was conducted with five colleagues, in order to test the online surveys and data extraction. The same SPSS syntax was used for the analysis to ensure reproducibility, and to allow any reader to verify what has been done.

RESULTS

Sample characteristics

Of 233 individuals who logged onto the YA survey site and 57 individuals who logged onto the parent survey site, 152 (65.2%) and 42 (73.7%) respectively provided both consent and complete data and were included in the analysis.

Young adult questionnaire results

The demographic information provided by the study participants is shown in Table 3. The majority of participants (n=142; 93.4%) were variant-positive, and among those 6.3% (n=9) had been diagnosed with cancer since having their PST. The number of females who completed the English questionnaire (PEQ) was significantly higher when compared to men (134/140, 95.7% versus 7/11, 63.6%; p=.003, Fisher's exact test). Among the PEQ the majority were variant-positive (96.9%) while in the Italian sample (PIQ) there were 19 (76.0%) variant-positive and six (24.0%) variant-negative respondents (p=.001, Fisher's exact test).

Finding out about their risk

Participants declared they received the information about their risk for the first time when between 5-30 years of age (20.0±5.6): 111 (75.5%) were informed after their 18th birthday, while 36 (24.5%) were informed earlier. Fifty-four YA participants (35.5%) were told by their mother, 19 (12.5%) by their father, 16 (10.5%) by both parents together, seven (4.6%) by their sister, 24 (15.8%) by other relatives such as aunts or cousins, and 26 (17.1%) by a person outside the family such as a genetic counsellor or a physician. Three participants (2.0%) had suspected they were at risk because of a family history of cancer and sought medical advice, and three (2.0%) reported the risk was openly discussed within their family. One-hundred and two participants (68.5%) reported they received the information at an unplanned time (75 in a face-to-face conversation and 27 in a telephone or social media call/message), while 43 (28.9%) received the information in a pre-planned conversation (38 in a face-to-face meeting and five in a telephone call). Two participants did not remember

how they received the information. The majority of participants (n=132; 86.8%) were told at that time that the tendency to cancer in their family could be due to a genetic change. A significant difference was observed in relation to the time taken for testing after disclosure: 73.1% of participants who had received the information from a person outside the family underwent PST within one year of obtaining the information ($\chi 2=19.951$, df=9, p=.018), compared to 41.7% of those told by a family member. Participants who were told about their potential genetic risk before their 18th birthday showed lower awareness of their risk (Q1, F1) ($Beta_{before18}$=*-.187,* R^2*=.028, p=.026*) and lower need for additional information (Q1, F2) ($Beta_{before18}$=*-.173,* R^2*=.037, p=.023*).

Decision-making process

The majority of participants (n=105; 75.5%) responded "myself" when asked about the person who decided that they would be tested, while "both myself and parents" was mentioned by 23 (16.5%), "parents" by four (2.9%), "aunt" by four (2.9%), and genetic counsellor/doctor by three (2.2%). The proportion of PEQ reporting the decision as made by themselves was significantly higher if compared to PIQ (96/116, 82.8% versus 9/23, 39.1%; $\chi 2=38.715$, df=4, p<.001). Participants who underwent PST within one year of obtaining the information were more likely to show proactivity (Q3, F1) than those who underwent PST between two and four years after (3.9±0.9 versus 3.2±1.3; F=2.987, p=.034). Becoming aware of potential genetic risk before age 18 or being informed by distant relatives predicted a lower perception of parents' pressure against testing (Q3 F2) ($Beta_{before18}$=-.220, $Beta_{other\text{-}relatives}$=-.617), unlike being tested between 18-25 years of age, that predicted higher perception

of parents disagreement about testing ($Beta_{before25}$=.260) (F(11,90)=2.028, p=.034). Men were found more likely than women to have undergone genetic testing because of their parent's decision both using variance analysis (3.4±1.3 versus 1.6±1.4; t(135)=4.640, p<.001) and multiple linear regression ($Beta_{female}$=.410). Pressure from parents toward testing was reported more frequently by participants without children than by those who had children (3.1±1.4 versus 2.5±1.4; t(134)=-2.771, p=.006), with a correlation confirmed by multiple linear regression ($Beta_{with\text{-}children}$=-.264). Also, participants who became aware of their risk before age 18 were less likely to undergo PST upon their parents' decision (Q3,F3) ($Beta_{before18}$=-.183) (F(11,110)=4.368, p<.001). Pressure by parents was more frequently reported by participants tested between 18-25 years than by those undergoing genetic testing between 26-30 years of age (3.0±1.4 versus 2.5±1.4; t(134)=2.202, p=.029).

Genetic test result

Respondents to the English questionnaire were more likely than PIQ to experience negative feelings (2.9±0.9 *versus* 2.4±1.1; t(131)=2.596, p=.011), and to worry for relatives (2.8±0.9 *versus* 2.2±0.9; t(133)=2.557, p=.012). Participants who had received the information on their risk in an unplanned conversation/call were more likely to experience negative feelings about their genetic test result (3.0±0.8 *versus* 2.7±0.9; t(125)=2.060; p=.041). A positive test result significantly predicted higher frequency of negative feelings about test outcome (Q4,F1) ($Beta_{gene\text{-}found}$ =.321) (F(11,111)=2.939, p=.002). Moreover, participants who received a positive test result were more likely to worry about their relatives (2.7±0.9 *versus* 1.8±0.8; t(133)=3.316, p=.001); ($Beta_{gene\text{-}found}$ =.213). Also having

children was a significant predictor of worrying about relatives (Q4,F4) ($Beta_{with\text{-}children}$=.287), (F(11,113)=2.098, p=.026). Having being diagnosed with cancer significantly reduced the perception of the test as helpful (Q4,F5) ($Beta_{cancer}$=-.198) (F(11,111)=3.072, p=.001).

Living with genetic risk

Italian participants were more likely to perceive the influence of lifestage (2.2±1.1 versus 1.4±1.1; t(127)=-2.701, p=.008), as well as those who were diagnosed with cancer ($Beta_{cancer}$=.289). Conversely, having children predicted a lower perception of lifestage influence (Q5,F1) ($Beta_{with\text{-}children}$=-.285) (F(11,108)=3.211, p=.001). Participants who received a positive test result were significantly more likely to perceive it as helpful to their own prevention and for relatives than those who received a negative test result (3.0±0.8 versus 1.7±1.2; t(121)=3.343, p=.001) ($Beta_{gene\text{-}found}$=.332, F(11,102)=1.956, p=.041); this was also true for participants who underwent PST between 18-25 years of age (2.0±0.7 versus 2.2±0.8; t(121)=2.127, p=.035) when compared to those tested at 26-30 years. Consistently, participants who received a positive test result were more likely to feel anxious than those who received a negative test result (2.2±0.7 versus 1.2±0.7; t(125)=3.043, p=.003) ($Beta_{gene\text{-}found}$=-.237, p=.012) (F(11,107)=2.246, p=.017).

Parent questionnaire results

The demographic information provided by the study participants is shown in Table 4. The majority of participants (n=25, 59.5%) had been previously diagnosed with cancer and 37 (88.1%) declared that there was a genetic tendency to cancer on their side of the family. Among those, 35 (94.6%)

had a genetic test at 47.4±6.2 age of years. Ten (23.8%) of those who had a PST had never had cancer.

Telling your children

All participants reported that they had told their children about the family risk themselves; the age of the children when told ranged from 5-44 years (21.8±6.6). The majority (n=28, 66.7%) decided to disclose the information in a planned conversation with their child(ren), eight (19.0%) told them in a casual way, and six (14.3%) took advantage of a moment when the child raised the issue. Concerning parents' reasons for telling their children about the family cancer risk, it was observed that participants who underwent genetic testing after having cancer were more likely to worry about the emotional impact on the child than those who underwent it before having cancer (2.3±1.1 versus 0.8±0.8; F=2.944, p=.050). However, participants who communicated the family cancer risk in a casual way to their children were less likely to have difficulties in communicating genetic status than those who planned a conversation with them (2.3±1.7 versus 1.0±1.1; F=4.164, p=.025). Consistently, a significant difference was found between participants with a genetic tendency to cancer in their partner's side of the family and participants with a genetic tendency in their own side of family: the first group were less likely to have difficulties in communicating genetic status (2.4±1.1 versus 0.7±0.9; t(23)=3.952, p=.001).

Children's experience of the PST

The majority (n=38, 94.7%) of parent participants told their children about the possibility of having a PST. Parents reported that the request for PST was made by the adult child themselves in 28 cases (73.7%), by the child

with one or both of the parents in five cases (13.2%), by the respondent or his partner in four cases (10.5%), and by the doctor in one case (2.6%).

Parents' feelings about PST for their children

Guilt about the possibility that the mutation might be inherited by their children (parent questionnaire, question 80 in suppl. file) was more common in the mothers ($Beta_{mother}$=.349 R^2.122). (F(1,34)=4.722, p=.037). However, all participants felt their children should be tested. The majority (n=26, 74.3%) also felt they had control over the decision their child made about the test.

DISCUSSION

The findings of this study suggest that young adults were told about the potential genetic risk at a mean age of 20±5.6 years. This is older than the age of 13.5±2.6 years in the American sample described by Tercyak et al.[24] and in general in the systematic review[15], where about half were informed before the age of 18 years old and all before 21 years of age. However, no YA was younger than 12 years of age when informed[15]. In contrast, in our sample the large majority (75.5.%) were informed after their 18th birthday. The large majority (68.5%) received the information in an unplanned conversation and only 2% of our sample reported that genetic risk was openly discussed in their family. We did not collect data on the age at which parents were tested, so were not able to compare the age at which the child was informed with the parents at which parents received their own test result, but this would be interesting to study in future. Informal discussion about their potential genetic risk was preferred by young people described by Metcalfe et al.[25] and in our sample parents were less likely to have difficulties in communicating genetic risk when it

happened in a casual way, as well as when they communicated the genetic risk of their partner's side of the family. However, YA who were told about their genetic risk in an unplanned situation were more likely to report negative feelings about their genetic test result. It could be hypothesized that a communication perceived by a YA to be 'casual' may hamper the full understanding of the risk, thus increasing the chance of a negative emotional impact. The majority of parents reported that they disclosed the information in a planned conversation, while the majority of YA reported that discussions were not usually planned, and due to anonymity of participants, we were not able to determine if participants (both YA and parents) belonged to the same families. In any case, it may be that a conversation that was planned by parents may have appeared unplanned to their children. The fact that parents made the decision to disclose without involving health professionals is concerning as Borry et al.[10] reported that parents were not able to transmit accurate information to their children regarding their genetic risk. It is possible that parents have not perceived the existence of support from genetic counsellors, even though Metcalfe et al.[26] showed that health professionals are increasingly being asked for advice from parents about risk disclosure to their children. However, reluctance by parents to involve health professionals may be partly due to the parents' wish to undertake this task alone[26,27]. While a previous systematic review[15] suggested that positive and negative emotional outcomes were not correlated with test results, our participants who received a variant-positive test result were more likely to experience negative feelings. Although the majority of the requests for genetic testing were made by YA offspring, the majority of parent participants felt they

had control over the decision their child made about the test and all felt their children should be tested, which is in line with previous findings, where parents appeared to have exerted pressure on their children during the decision making process about testing[15]. These issues raise the ethical problem of how health professionals can respect young adults' developing autonomy[28–32]. Werner-Lin et al.[32] investigated genetic counsellors' perspectives on counselling clients aged between 18-25 years, using an online survey: a primary challenge reported was navigating family dynamics in counselling sessions. However, our findings show that YA who were strongly influenced by their parents to be tested were less likely to feel anxious. This result may confirm that YA did not completely understand the implications of the genetic test but complied because of parental pressure, and potentially felt relieved of the responsibility to make their own decisions. An American study indicated that the current generation of YA have higher levels of student debt and are more likely to experience poverty and unemployment, while 53% of emerging adults aged 18-24 years currently lived with parents[33,34]. This is also true in Italy, where 62.5% of YA aged 18-34 years live with their nuclear families[35,36]. Living independently is one of the key developmental tasks of emerging adulthood[37]. If YA are co-resident with their parents, this could slow down the process of achieving autonomy as an adult. It is reasonable to hypothesise that this style of life has an impact on developmental tasks, reducing the autonomy of YA in their decision making. In fact, in our sample the number of PIQ who had been tested based on their own decision was significantly lower if compared to PEQ. However, genetic counsellors may have a responsibility to enable young

people to challenge decisions made by their parents that may be inappropriate for them[38]; it may be that parents do not always make the best possible decision for their offspring, but usually one that is intended to support them. In the context of PST, where there is uncertainty about the potential harm and/or benefits, Cohen believes that the parent's decision should prevail over their offspring's decision[39]. However, with regard to the principle of decision-making by a surrogate Buchanan and Brock[40] provided data on the fact that there may be a failure by parents to make a decision in the best interests of their children. The evidence of this study highlights the need for a comprehensive, longitudinal counselling process with appropriate timing and setting, which supports 'parent-to-offspring' risk communication first and YA's decision making about PST and risk management afterwards. This would include emphasising that disclosure of genetic risk is a gradual and dynamic process in the family, and where children are told at an early age, this should be followed with further age-appropriate information.

Strengths and limitations

The limited number of PIQ reduced the possibility of observing differences between groups about their experience of PST. This could be the result of difficulties in recruiting: only 39.3% of PIQ had accessed the questionnaire via the Internet, compared to 100% of PEQ. Another reason could be less interest in the Italian population regarding sharing information on medical issues via the Internet. Furthermore, the possibility of generalizing the results of factor analysis could be hampered by the small sample size[41–45], particularly for the parent questionnaire. Moreover, almost all participants were variant-positive. It may be that potential participants who received

negative test results were no longer sufficiently interested in the topic to respond, or perceived that the topic was not relevant to them. Additionally, another limitation could be that data were collected retrospectively and not at the time of PST. Moreover, the choice of statistical tests and the SPSS outcomes were assessed by all the authors, who are experienced researchers, to maximise the validity of the analysis.

Conclusions

In conclusion, there is much research to do on this topic, and the results presented here need to be more fully explored. However, the findings of this study could contribute to improving clinical practice. They indicate a need both for publicising the supportive and educational role of genetic services. It is therefore important to emphasise that young adults may benefit from a multistep approach for undergoing genetic testing, and parents need to be more informed that genetic counselling is a place where information is obtained and young adults can freely talk about the decision, regardless of whether they want to be tested or not.

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CONFLICT OF INTEREST

The authors have no conflicts of interest.

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FIGURE 1: INCLUSION AND EXCLUSION CRITERIA

FIGURE 2 : RECRUITMENT FLOW-CHART

TABLE 1: FACTORS FOR EACH QUESTION ANALYSED

TABLE 2 (A-E): MULTIPLE LINEAR REGRESSION ANALYSIS OF FACTORS IDENTIFIED BY EFA

TABLE 3: SAMPLE CHARACTERISTICS: YOUNG ADULT PARTICIPANTS

TABLE 4: SAMPLE CHARACTERISTICS: PARENT PARTICIPANT

FIGURE 1: INCLUSION AND EXCLUSION CRITERIA

Participants were *eligible* to take part in the study fitted either of the two groups below.

1. Young adults who were:
    - aged 18-30 years when they underwent the presymptomatic genetic test for a familial cancer syndrome
    - without personal history of cancer when they underwent a presymptomatic genetic test and
    - members of families with a hereditary cancer predisposition.
2. Parents of young adults who were tested between 18-30 years of age.

Individuals in either group were *ineligible* if they were unable:

- to provide informed consent due to mental incapacity or active psychotic illness or
- unable to complete a survey in either English or Italian.

FIGURE 2 : RECRUITMENT FLOW-CHART

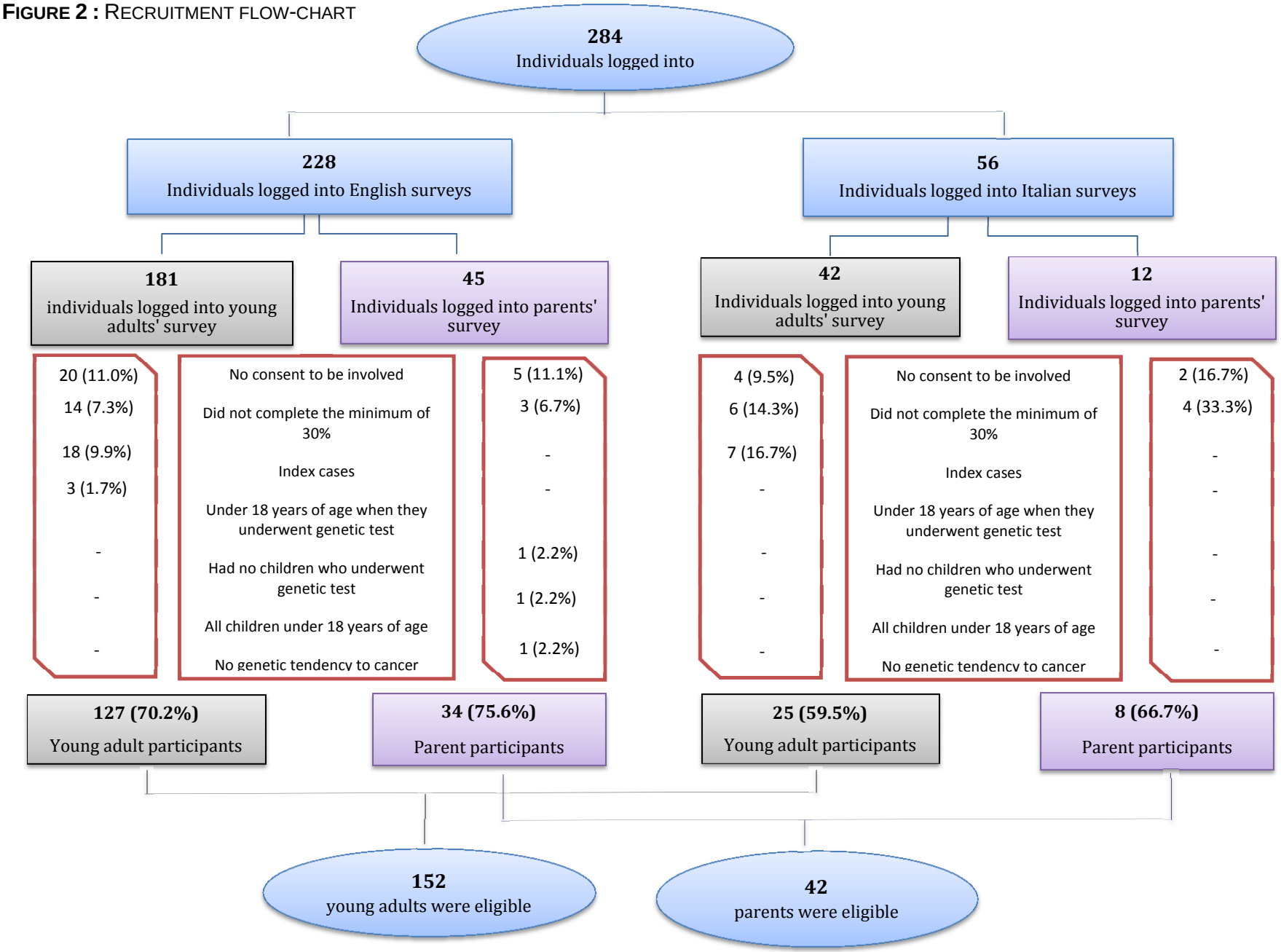

TABLE 1: FACTORS FOR EACH QUESTION ANALYSED

Q1: How did you react to the news that there might be a genetic condition in your family?"

	STRONGLY OR SOMEWHAT DISAGREE	NEITHER AGREE NOR DISAGREE	STRONGLY OR SOMEWHAT AGREE
Factor 1: Awareness			
I did not know what it really meant	31 (20.7%)	15 (10.0%)	104 (69.3%)
I looked for information online	10 (6.8%)	15 (10.1%)	123 (83.1%)
I was more conscious of my risk	36 (23.8%)	20 (13.2%)	95 (62.9%)
I arranged the first counselling session to have a genetic blood test	34 (54.0%)	24 (16.0%)	45 (30.0%)
I felt it explained things I had been wondering about	43 (28.9%)	46 (30.9%)	60 (40.3%)
Factor 2: Need for information			
I arranged the first counselling session to discuss my risk	81 (22.8%)	13 (8.7%)	102 (68.5%)
I wanted to know some more about it at the time	22 (14.7%)	16 (10.7%)	112 (74.7%)

Q2: How did you feel about the genetic counselling?

	STRONGLY OR SOMEWHAT DISAGREE	NEITHER AGREE NOR DISAGREE	STRONGLY OR SOMEWHAT AGREE
Factor 1: Satisfaction with genetic counselling			
The doctor or genetic counsellor showed an interest in your personal situation regarding the cancer family history	14 (9.6%)	10 (6.8%)	123 (83.7%)
The doctor or genetic counsellor explained your risk to you clearly	12 (8.2%)	8 (5.4%)	123 (83.1%)
The doctor or genetic counsellor met your expectations of him or her	19 (13.0%)	14 (9.6%)	113 (77.4%)
The doctor or genetic counsellor treated you as an individual	15 (10.2%)	10 (6.8%)	122 (83.0%)
You would be comfortable in calling the doctor or genetic counsellor to ask further questions	27 (18.4%)	16 (10.9%)	104 (70.7%)
The doctor or genetic counsellor listened to what you had to say	14 (9.7%)	15 (10.3%)	116 (80.0%)
The doctor or genetic counsellor was considerate of your emotional state during the meeting	20 (13.7%)	16 (11.0%)	110 (75.3%)

You are satisfied with the way that information was communicated to you	21 (14.3%)	11 (7.5%)	115 (78.3%)
The doctor or genetic counsellor understood what was really concerning you	21 (14.5%)	14 (24.1%)	110 (75.8%)
The doctor or genetic counsellor made you feel you were "in good hands"	22 (15.1%)	14 (9.6%)	110 (75.3%)
The doctor or genetic counsellor made you feel that they knew how to handle situations like your's	23 (15.6%)	15 (10.2%)	109 (74.1%)
The doctor or genetic counsellor gave you enough of their time	16 (10.9%)	13 (8.8%)	118 (80.3%)
The doctor or genetic counsellor was sensitive and tactful during your conversation	18 (12.2%)	8 (5.4%)	121 (82.3%)
The doctor or genetic counsellor seemed to be an expert in the field	19 (12.9%)	11 (7.5%)	117 (79.6%)
The doctor or genetic counsellor helped you deal with any concerns you had	19 (13.0%)	20 (13.7%)	107 (73.3%)
You felt comfortable to talk about yourself during the genetic counselling session	17 (11.6%)	16 (10.9%)	114 (77.5%)
You were satisfied with the length of time you had to wait until your first appointment	33 (22.6%)	19 (13.0%)	94 (64.4%)
You were satisfied with the information your received during the genetic counselling appointment	22 (15.0%)	11 (7.5%)	114 (77.5%)
If a friend needed similar help you would recommend this clinic to him or her	19 (13.0%)	18 (12.2%)	110 (74.8%)
The counselling was given in an appropriate setting	10 (6.8%)	11 (7.5%)	126 (85.7%)
Overall you are satisfied with the genetic counselling service	18 (12.2%)	14 (9.5%)	115 (78.3%)

Q3: What were your reasons for wanting to be tested?

	STRONGLY OR SOMEWHAT DISAGREE	NEITHER AGREE NOR DISAGREE	STRONGLY OR SOMEWHAT AGREE
Factor 1: Proactivity			
I wanted to try to help advance research	20 (14.5%)	26 (18.8%)	87 (63.0%)
I wanted to know if I need to get cancer screening tests more often	4 (2.9%)	7 (5.1%)	113 (83.1%)
I wanted to be reassured	9 (6.5%)	29 (21.0%)	92 (66.7%)
I wanted to make a decision about surgery to reduce my risk	13 (9.4%)	14 (10.1%)	97 (70.5%)
I made my own decision	5 (3.6%)	8 (5.8%)	95 (68.8%)

My decision was influenced by family experience	14 (10.2%)	21 (15.3%)	88 (67.7%)
Factor 2: Parents' pressure against testing			
My mother warned me about having the test	78 (60.0%)	26 (19.0%)	16 (11.7%)
My father warned me about having the test	88 (64.8%)	25 (18.4%)	8 (5.9%)
My mother advised me to wait, but I decided to have it	95 (68.8%)	16 (11.6%)	9 (6.5%)
My father advised me to wait, but I decided to have it	100 (74.8%)	15 (11.1%)	5 (3.8%)
Factor 3: Parents' decision to be tested			
I had genetic testing because of pressure from my family members	99 (71.8%)	19 (13.8%)	16 (11.6%)
I had genetic testing because my parent asked me to do it	92 (66.7%)	23 (16.7%)	17 (12.3%)
Factor 4: Concern for children			
I wanted to learn about my children's risk or risks to any children I may have	10 (7.3%)	19 (13.9%)	92 (67.1%)
I wanted to make a decision about having (more) children	34 (24.8%)	26 (19.0%)	59 (43.2%)
Factor 5: Parent's pressure for testing			
My mother strongly encouraged me	35 (25.3%)	31 (22.5%)	59 (42.7%)
My father strongly encouraged me	44 (32.3%)	36 (26.4%)	44 (32.3%)

Q4: How did you feel after receiving your genetic test result?

	STRONGLY OR SOMEWHAT DISAGREE	STRONGLY OR SOMEWHAT AGREE
Factor 1: Negative feelings		
I felt upset about my test result	36 (26.7%)	99 (73.3%)
I felt sad about my test result	27 (20.0%)	108 (80.0%)
I felt anxious or nervous about my test result	40 (29.6%)	95 (70.4%)
I was more worried about my risk of getting cancer	28 (20.7%)	107 (79.2%)
I felt a loss of control	70 (51.5)	66 (48.5%)
Factor 2: Negative impact on relationships		
I felt guilty about my test result	95 (69.8%)	41 (30.1%)
I had problems enjoying life because of my test result	84 (62.2%)	51 (37.5%)
I was worried other people might discuss this behind my back	114 (85.1%)	20 (14.9%)

I was worried other people might think less of me because of my result	114 (83.8%)	22 (16.2%)
I felt more distant from family members	115 (84.6)	21 (15.4%)
Factor 3: Uncertainties about the meaning of test result		
I was uncertain about what my test result meant for my cancer risk	112 (82.4%)	24 (17.6%)
I was uncertain about what my test result meant for my children or any children I may have	103 (76.8%)	31 (23.2%)
I was uncertain about what my test result meant for my family's cancer risk	106 (78.5%)	29 (21.5%)
Factor 4: Worry for relatives		
I was worried because of the possibility of passing the mutation to my children or any children I may have	23 (17.0%)	112 (83.0%)
I felt guilty about my family	87 (64.0%)	49 (36.0%)
Factor 5: Perceiving the test as helpful		
I felt relieved about my test result	83 (61.5%)	52 (38.5%)
I felt able to plan my future	42 (31.1%)	93 (68.9%)

Q5: How did you feel living with your genetic risk?

	STRONGLY OR SOMEWHAT DISAGREE	STRONGLY OR SOMEWHAT AGREE
Factor 1: Influence on lifestage perception		
I have wondered about when to share my genetic risk with a new partner	46 (35.4%)	42 (32.4%)
I have wondered about how early in a relationship to discuss having children	47 (35.9%)	37 (28.2%)
I have wondered about how early in a relationship to discuss surgery to reduce my risk	46 (35.1%)	42 (32.0%)
I try not to think about the cancer risk because I am too young yet for screening	69 (53.1%)	32 (24.6%)
Factor 2: Impact of test result on own prevention and on relatives		
Having time before the regular cancer screening was due to start gave me the opportunity to think about it	26 (20.1%)	86 (66.7%)
Having time before the cancer screening was due to start gave me the opportunity to think about having surgery to reduce my risk	29 (11.6%)	95 (73.6%)
I understood my choice for cancer prevention or early detection clearly	10 (7.8%)	111 (87.7%)
I felt frustrated that there are no ways I can completely prevent cancer	32 (24.4%)	89 (68.0%)

I felt satisfied with family communication about my genetic test result	24 (18.7%)	96 (74.4%)
I was worried about the possibility of my children (or any children I may have) getting cancer	12 (9.3%)	103 (79.3%)
I was feeling guilty about possibly passing on the disease risk to my children or any children I may have	22 (16.8%)	92 (70.3%)
Factor 3: Anxiety		
I was having difficulty making decisions about cancer screening or measures to reduce my risk	89 (68.4%)	33 (25.4%)
I thought about having risk -reducing surgery sooner rather than later	27 (20.7%)	90 (69.3%)
My parents strongly encouraged me to have surgery to reduce my risk of cancer	78 (59.5%)	30 (23.0%)
Thinking about my test result has affected my work or family life	58 (44.3%)	66 (50.4%)
I had difficulty talking about my test results with family members	95 (73.8%)	30 (23.8%)
I decided to limit the number of children I have because I may pass on the mutation	70 (53.5%)	40 (30.6%)
I feel anxious waiting for the first or next screening	29 (22.4%)	83 (63.8%)
Factor 4: Protection of self and children		
I regretted my choice to have children	73 (56.6%)	12 (9.4%)
I try to do all I can to stay alive for my children	6 (4.6%)	75 (58.2%)
I have confidence in the cancer screening procedures	28 (21.6%)	95 (73.8%)

Q6: What were your reasons for telling or not telling your children about the family cancer risk?

	STRONGLY OR SOMEWHAT DISAGREE	NEITHER AGREE NOR DISAGREE	STRONGLY OR SOMEWHAT AGREE
Factor 1: Making children aware			
I wanted to provide access to information for my children	4 (10.0%)	2 (5.0%)	34 (85.0%)
I wanted to make my children aware of the risk	4 (10.0%)	2 (5.0%)	34 (85.0%)
I wanted to share my genetic test results with my children so they could be tested	4 (10.3%)	1 (2.6%)	36 (90.0%)
I wanted to explain the family history of cancer	4 (10.3%)	3 (7.7%)	32 (80.0%)
I wanted to share my genetic test results with my children because of my grandchildren or future grandchildren	4 (10.0%)	6 (15.0%)	27 (67.5%)

I felt it was the appropriate age to tell them	5 (12.8%)	4 (10.3%)	30 (76.9%)
I wanted them to be able to have screening	4 (10.5%)	1 (2.6%)	32 (88.9%)
I thought my children were too young to know	22 (61.1%)	1 (2.8%)	3 (8.3%)
I was not ready to share the news	26 (70.3%)	2 (5.4%)	4 (10.8%)
There was no medical reason to tell them	28 (77.8%)	1 (2.8%)	1 (2.8%)
Factor 2: Worry about emotional impact on children			
I thought it might make my children anxious	16 (43.2%)	7 (18.9%)	10 (27.8%)
I thought it might increase my children's fear of getting cancer	19 (51.4%)	3 (8.1%)	11 (29.7%)
I thought it might increase my children's worry about my and my partner's health	16 (43.2%)	7 (18.9%)	10 (27.0%)
I am still coping with the test results	21 (56.8%)	3 (8.1%)	8 (21.6%)
Factor 3: Difficulties in communicating own genetic status			
I wanted to share my partner's genetic test results with my children so they could be tested	2 (5.6%)	1 (2.8%)	9 (22.5%)
I wanted to share my partner's genetic test results with my children because of my grandchildren or future grandchildren	2 (5.6%)	1 (2.8%)	9(25.0%)
I didn't intend to tell them but they accidentally found out	19 (52.8%)	1 (2.8%)	1 (2.8%)
I thought it was unnecessary to make my children aware of the family history	27 (73.0%)	1 (2.7%)	4 (10.8%)

TABLE 2 (A): MULTIPLE LINEAR REGRESSION ANALYSIS OF FACTORS IDENTIFIED BY EFA

	Q1: How did you react to the news that there might be a genetic condition in your family?"						*Q2: How did you feel about the genetic counselling?*		
	Factor 1: Awareness			**Factor 2: Need for information**			**Factor 1: Satisfaction with genetic counselling**		
	B	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***
Gender	-.224	1.791	-.012	*-.331*	*.931*	*-.033*	-.685	9.599	-.007
Age	-.116	.084	-.152	*-.022*	*.043*	*-.055*	-.247	.464	-.062
Having children	-.896	.909	-.103	*-.139*	*.459*	*-.031*	4.207	4.942	.092
Age at PST	.059	.814	.007	*.308*	*.418*	*.069*	6.315	4.429	.137
PST result	2.871	1.770	.147	*.834*	*.918*	*.083*	8.912	9.440	.087
Having cancer	1.184	1.567	.068	*-.783*	*.772*	*-.092*	-14.680	8.349	-.162
YA told by first-degree relatives	-4.616	2.130	-.514*	*-1.259*	*1.107*	*-.274*	-4.446	11.423	-.093
YA told by distant relatives	-4.276	2.329	-.358	*-.672*	*1.211*	*-.109*	-6.356	12.542	-.100
YA told by members outside the family	-3.077	2.153	-.272	*-.945*	*1.116*	*-.164*	7.183	11.552	.119
How YA received the information	-.035	.924	-.004	*-.586*	*.479*	*-.120*	-.363	5.146	-.007
Age at information received were entered into YA models	-2.174	.910	-.211*	*-1.013*	*460*	*-.195**	-5.375	4.755	-.102
	F(11,121)=1.587, p=.111			*F(11,125)=1.144, p=.333*			*F(11,119)=1.275, p=.247*		

TABLE 2 (B): MULTIPLE LINEAR REGRESSION ANALYSIS OF FACTORS IDENTIFIED BY EFA

	Q3: What were your reasons for wanting to be tested?														
	Factor 1: Proactivity			**Factor 2: Parents' pressure against testing**			**Factor 3: Parents' decision to be tested**			**Factor 4: Concern for children**			**Factor 5: Parent's pressure for testing**		
	B	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***
Gender	1.313	1.215	.128	*-2.335*	*1.784*	*-.141*	-.4.303	.947	-.410**	.104	.993	.011	-2.412	1.252	-.194
Age	.059	.066	.123	*-.046*	*.085*	*-.074*	.056	.044	.130	-.082	.049	-.222	-.019	.058	-.040

Having children	-1.113	.671	-.215	*-.600*	*.850*	*-.084*	-1.288	.480	-.264**	.689	.516	.159	-1.104	.592	-.210
Age at PST	-.214	.607	-.041	*1.867*	*.761*	*.260**	1.022	.432	.208*	.567	.467	.130	.943	.538	.178
PST result	.439	1.328	.039	*.734*	*1.765*	*.044*	.067	.869	.007	.792	.964	.085	1.382	1.123	.121
Having cancer	1.694	1.106	.177	*2.827*	*1.469*	*.200*	.886	.839	.090	1.501	.800	.195	.208	.966	.021
YA told by first-degree relatives	-2.678	1.548	-.509	*-4.220*	*2.180*	*-.572*	1.002	1.087	.199	-.310	1.188	-.070	3.164	1.567	.586*
YA told by distant relatives	-2.385	1.685	-.364	*-5.910*	*2.301*	*-.617**	.379	1.194	.057	.459	1.287	.078	1.871	1.637	.267
YA told by members outside the family	-1.538	1.541	-.224	*-4.075*	*2.206*	*-.435*	.615	1.100	.095	.354	1.210	0.63	3.670	1.576	.546*
How YA received the information	-.372	.708	-.065	*-.158*	*.910*	*-.020*	-.053	.515	.010	.059	.553	.012	.111	.631	.019
Age at information received were entered into YA models	.793	.656	.133	*-1.875*	*.881*	*-.220**	-1.048	.480	-.183*	-.375	.509	-.074	-1.358	.598	-.222*
	F(11,76)=1.610, p=.113			*F(11,90)=2.028, p=.034*			*F(11,110)=4.368, p<.001*			*F(11,93)=1.375, p=.198*			*F(11,94)=2.606, p=.006*		

TABLE 2 (C): MULTIPLE LINEAR REGRESSION ANALYSIS OF FACTORS IDENTIFIED BY EFA

	Q4: How did you feel after receiving your genetic test result?														
	Factor 1: Negative feelings			**Factor 2: Negative impact on relationships**			**Factor 3: Uncertainties about the meaning of test result**			**Factor 4: Worry for relatives**			**Factor 5: Perceiving the test as helpful**		
	B	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***
Gender	2.878	1.638	.167	*1.456*	*1.345*	*.112*	.213	1.035	.020	-.279	.729	-.037	-.492	.697	-.066
Age	.044	.081	.061	*.013*	*.070*	*.023*	-.032	.052	-.072	-.056	.037	-.176	-.033	.036	-.105
Having children	-.926	.898	-.109	*-.635*	*.754*	*-.099*	.592	.578	.115	1.064	.401	.287**	.751	.383	.205
Age at PST	.405	.794	.048	*.056*	*.652*	*.009*	.571	.505	.111	.574	.353	.154	-.055	.338	-.015
PST result	5.891	1.719	.321**	*.855*	*1.308*	*.066*	-.338	1.005	-.033	1.612	.710	.213*	1.204	.722	.153
Having cancer	-.518	1.458	-.032	*1.179*	*1.209*	*.096*	3.137	.928	.319*	1.255	.655	.175	-1.392	.623	-.198*
YA told by first-degree relatives	1.441	2.002	.166	*.466*	*1.659*	*0.72*	.465	1.273	.089	-.149	.899	-.039	-.233	.930	-.062

YA told by distant relatives	1.358	2.189	.120	*1.190*	*1.812*	*.140*	.074	1.394	.011	-.232	.984	-.047	-.457	.990	-.094
YA told by members outside the family	-.447	2.004	-.041	*.791*	*1.659*	*.098*	1.004	1.271	.155	.393	.898	.084	-1.842	.934	-.399
How YA received the information	.790	.938	.084	*-.176*	*.778*	*-.025*	.236	.600	.042	.163	.421	.040	-.320	.403	-.079
Age at information received were entered into YA models	-.249	.866	-.025	*-.651*	*.730*	*-.086*	-.014	.554	-.002	-.203	.389	-.524	-.207	.370	-.049
	F(11,111)=2.939, p=.002			*F(11,111)=.640, p=.791*			*F(11,111)=1.561, p=.120*			*F(11,111)=2.939, p=.002*			*F(11,111)=3.072, p=.001*		

TABLE 2 (D): MULTIPLE LINEAR REGRESSION ANALYSIS OF FACTORS IDENTIFIED BY EFA

	Q5: How did you feel living with your genetic risk?											
	Factor 1: Influence on lifestage perception			**Factor 2: Impact of test result on own prevention and on relatives**			**Factor 3: Anxiety**			**Factor 4: Protection of self and children**		
	B	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***	***B***	***SE B***	***Beta***
Gender	.058	1.765	.003	*-1.743*	*2.632*	*-.066*	4.030	2.089	.182	-.359	1.122	-.108
Age	-.073	.089	-.094	*-.081*	*.123*	*-.081*	-.026	.105	-.030	-.070	.057	-.138
Having children	-2.594	.979	-.285**	*1.372*	*1.351*	*.116*	-.660	1.162	-.064	2.956	.621	.498**
Age at PST	1.114	.847	.121	*2.268*	*1.177*	*.190*	1.509	1.005	.144	.279	.540	.047
PST result	1.671	1.867	.080	*9.587*	*2.899*	*.332***	5.630	2.215	.237*	1.000	1.189	.074
Having cancer	5.002	1.544	.289**	*2.754*	*2.116*	*.125*	3.607	1.830	.184	2.989	.982	.266**
YA told by first-degree relatives	.253	2.315	-.027	*-.135*	*3.163*	*-.011*	-.735	2.744	-.069	-1.658	1.472	-.273
YA told by distant relatives	-.215	2.463	-.018	*-.230*	*3.386*	*-.015*	.491	2.916	.036	-1.452	1.576	-.183
YA told by members outside the family	-1.331	2.312	-.117	*-1.719*	*3.182*	*-.115*	-.752	2.757	-.056	-1.842	1.476	-.245

How YA received the information	-1.656	1.050	-.162	*-2.429*	*1.484*	*-.183*	-.319	1.258	-.027	.068	.683	.010
Age at information received were entered into YA models	.082	.925	.008	*.344*	*1.297*	*.025*	-.497	1.110	-.041	-.299	.596	-.043
	F(11,108)=3.211, p=.001			*F(11,102)=1.956, p=.041*			*F(11,107)=2.246, p=.017*			*F(11,107)=3.815, p<.001*		

TABLE 2 (E): MULTIPLE LINEAR REGRESSION ANALYSIS OF FACTORS IDENTIFIED BY EFA°

	Q6: What were your reasons for telling or not telling your children about the family cancer risk?					
	Factor 1: Making children aware			**Factor 2: Worry about emotional impact on children**		
	B	***SE B***	***Beta***	***B***	***SE B***	***Beta***
Gender	-7.238	14.655	-.260	*-6.308*	*3.818*	*-.496*
Age	-.087	.846	-.089	*-.249*	*.284*	*-.476*
Having cancer	8.872	11.184	.478	*3.037*	*2.744*	*.356*
Age at PST	-.207	.907	-.177	*-.001*	*.336*	*-.003*
Being the first person tested in the family	1.169	9.395	.063	*1.096*	*2.736*	*.126*
Way of communication to children	3.632	10.494	.171	*1.289*	*2.396*	*.144*
PST requested by children	15.562	11.476	.784	*-1.813*	*3.069*	*-.196*
PST requested by parents	18.256	13.078	.774	*-4.791*	*3.690*	*-.377*
	F(8,7)=.451, p=.856			*F(8,15)=.951, p=.527*		

**p<.05*
***p<.01*
°*Multiple linear regression was not performed for factor 3 because the assumptions*

TABLE 3: SAMPLE CHARACTERISTICS: YOUNG ADULT PARTICIPANTS

	ALL (N=152)	PEQ§ (N=127)	PIQ§ (N=25)	*p-value*
Age at questionnaire (years)				
mean±SD	29.5±5.6	29.6±5.9	28.7±3.7	0.463 [a]
Age at PST (years)				
mean±SD	24.7±3.7	24.7±3.8	25.0±3.4	0.700 [a]
Gender				
Male	11 (7.2%)	2 (1.6%)	9 (36.0%)	0.000[b,*]
Female	140 (92.1%)	124 (97.6%)	16 (64.0%)	
I prefer not to say	1 (0.7%)	1 (0.8%)	0	
Country				
Italy	25 (16.4%)	0	25 (100%)	-
United Kingdom	63 (41.4%)	63 (49.6%)	0	
United States of America	47 (30.9%)	47 (37.0%)	0	
Other countries	17 (11.2%)	17 (13.4%)	0	
Education				
Primary school	1 (0.7%)	1 (0.8%)	0	0.191[b]
Secondary school	15 (9.9%)	15 (11.8%)	0	
Post-secondary educ.	49 (32.2%)	38 (29.9%)	11 (44.0%)	
University degree	62 (40.8%)	50 (39.4%)	12 (48.0%)	
Postgraduate degree	25 (16.4%)	23 (18.1%)	2 (8.0%)	
Daily work				
Paid employment	112 (73.7%)	94 (74.0%)	18 (72.0%)	0.176[b]
Voluntary employment	2 (1.3%)	1 (0.8%)	1 (4.0%)	
Student	18 (11.8%)	13 (10.2%)	5 (20.0%)	
Homemaker	15 (9.9%)	15 (11.8%)	0	
Not working not student	5 (3.3%)	4 (3.1%)	1 (4.0%)	
Marital status				

Single (never married)	48 (31.6&)	33 (26.0%)	15 (60.0%)	0.009[b]
Married	67 (44.1%)	60 (47.2%)	7 (28.0%)	
Divorced	7 (4.6%)	6 (4.7%)	1 (4.0%)	
Living with a partner	30 (19.7%)	28 (22.0%)	2 (8.0%)	
Children				
Yes	73 (48.0%)	69 (54.3%)	4 (16.0%)	0.000[c]
No	79 (52.0%)	58 (45.7%)	21 (84.0%)	
Condition tested				
Cowden syndrome	1 (0.7%)			
Familial adenomatous polyposis	14 (9.2%)			
Hereditary breast and ovarian cancer	111 (73.0%)			
Lynch syndrome	26 (17.1%)			

[§] PEQ was used to indicate the participants responding to the English questionnaire and PIQ the participants responding to the Italian questionnaire

[*] "I prefer not to say" answer was excluded from the analysis

[a] Independent samples T-test

[b] Pearson chi-squared test

[c] Fisher's exact test

TABLE 4: SAMPLE CHARACTHERISTICS: PARENT PARTICIPANTS

		ALL (N=42)	PEQ (N=34)	PIQ (N=8)	*p-value*
Age at questionnaire (years)					
mean±SD		51.9±7.6	51.7±7.3	55.1±3.8	0.211 [a]
Gender					
	Male	4 (9.5%)	4 (100.0%)	0	0.572[c]
	Female	38 (90.5%)	30 (78.9%)	8 (21.1%)	
Country					
	Italy	8 (19.0%)	0	8 (100.0%)	-
	United Kingdom	17 (40.5%)	17 (50.0%)	0	
	United State of America	11 (26.2%)	11 (32.4%)	0	
	Other countries	6 (14.3%)	6 (17.6%)	0	
Education					
	Secondary school	10 (23.8%)	9 (26.5%)	1 (12.5%)	0.461[b]
	Post-secondary educat.	20 (47.6%)	15 (44.1%)	5 (62.5%)	
	University degree	7 (16.7%)	5 (14.7%)	2 (25.0%)	
	Postgraduate degree	5 (11.9%)	5 (14.7%)	0	
Daily work					
	Paid employment	29 (69.0%)	23 (67.6%)	6 (75.0%)	0.392[b]
	Homemaker	7 (16.7%)	5 (14.7%)	2 (25.0%)	
	Not working not student	6 (14.3%)	6(17.6%)	0	
Marital status					
	Single (never married)	1 (2.9%)	1 (2.9%)	0	0.378[b]
	Married	32 (76.2%)	24 (70.6%)	8 (100.0%)	
	Married	8 (19.0%)	8 (23.5%)	0	
	Living with a partner	1 (2.4%)	1 (2.3%)	0	
Condition tested					

	Cowden syndrome	4 (11.4%)
	Familial adenomatous polyposis	1 (2.9%)
	Hereditary breast and ovarian cancer	24 (68.6%)
	Lynch syndrome	6 (17.1%)

[a] Independent samples T-test

[b] Pearson chi-squared test

[c] Fisher's exact test