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J Health Psychol 2002 7: 491

DOI: 10.1177/1359105302007005628

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Predictive Genetic Testing and Beyond: A Theory of Engagement

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ACKNOWLEDGEMENTS. The author acknowledges the participation of the subjects with thanks, and is grateful for access to patients provided by Prof. D. G. R. Evans, Manchester; Dr C. McKeown, Birmingham; Dr J. Mackay, Cambridge; and Dr A. Ellis, Liverpool. This paper is based on research carried out by the author for her PhD at the Centre for Family Research, University of Cambridge, supervised by Prof. Martin Richards and Dr Nina Hallowell. The author is grateful for further comments on the manuscript provided by Dr Amy Silver. The Cancer Research Campaign supported this work (grant-holder: Prof. Martin Richards).

COMPETING INTERESTS: None declared.

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Journal of Health Psychology
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London, Thousand Oaks and New Delhi,
[1359-1053(200209)7:5]
Vol 7(5) 491-508; 024628

Abstract

This article presents a tentative grounded theory, which can provide some explanation of variation in behaviour around predictive genetic testing (PGT) for Hereditary Non-Polyposis Colorectal Cancer (HNPCC), based on interviews with individuals ($n = 55$) from families with a clinical diagnosis of HNPCC, 12 of whom were followed through the PGT protocol. The theory is built around a core category of engagement, a newly constructed concept reflecting the degree of cognitive and emotional involvement with cancer risk in individuals from these families, and models the psychosocial process of engaging with cancer risk. The degree of engagement at the time of testing can explain variations in approaches and reactions to PGT. A series of *social* factors, many related to the experiences of family life, emerged as either facilitating or blocking the process of engaging with cancer risk; a series of *psychological* factors emerged as interacting in a recursive, dynamic manner with each other and with engagement status. The degree of engagement can change with the unfolding of time and events in family life. The theory of engagement (TE) provides an explanatory framework for understanding behaviour related to PGT for HNPCC, and can potentially be applied to research on risk perception in the social sciences more generally. In addition, the theory may have potential uses in the genetics clinic, in identifying individuals at risk of adverse reactions to PGT for cancer, thus enabling better targeting of genetic counselling resources.

Keywords

engagement, genetic testing, grounded theory, HNPCC, risk perception

Introduction

PREDICTIVE GENETIC TESTING (PGT) is available for an increasing number of dominantly inherited disorders, including some inherited cancer predisposing syndromes. Recent studies have identified some factors that may influence decisions about whether or not to take such a test, such as gender (Bloch, Fahy, & Hayden, 1989; Dudok de Wit, 1997; Lerman, Schwarz, Lin, Hughes, Narod, & Lynch, 1997); family experience of the disease (Dudok de Wit, 1997; Lerman, Marshall, Audrain, & Gomez-Caminero, 1996; Lerman, Narod, Schulman, Hughes, Gomez-Caminero, Bonney, Gold, Trock, Main, Lynch, Fulmore, Snyder, Lemon, Conway, Tonin, Lenoir, & Lynch, 1996); higher levels of cancer-related worry (Glanz, Grove, Lerman, Gotay, & LeMarchand, 1999); severity of the condition (Evans, Maher, Macleod, Davies, & Craufurd, 1997); relief of uncertainty (Tibben, Duivenvoorden, Vegter-van der Vlis, Niermeijer, Frets, van de Kamp, Roos, Rooijmans, & Verhage, 1993; Tibben, Frets, van de Kamp, Niermeijer, Vegter-van der Vlis, Roos, Rooijmans, van Ommen, & Verhage, 1993); expectations about test results (Kessler, 1988, 1994; Kessler & Bloch, 1989) and wishing to clarify the position for children (Lynch, Lemon, Durham, Tinley, Connolly, Lynch, Surdam, Orinion, Slominski-Caster, Watson, Lerman, Tonin, Lenoir, Serova, & Narod, 1997).

Previous psychometric studies have not been found to consistently predict outcomes for PGT (Broadstock, Michie, & Marteau, 2000). However, case studies and interview studies have identified some complex family issues such as preselection and scapegoating (Kessler, 1988; Kessler & Bloch, 1989; Richards, 1996); survivor guilt (Biesecker, Boenke, Calzone, Markel, Garber, Collins, & Weber, 1993; Dudok de Wit, 1997; Lynch, Lemon, Karr, Franklin, Lynch, Watson, Tinley, Lerman, & Carter, 1997); family communication difficulties (Huggins, Bloch, Wiggins, Adam, Suchowersky, Trew, Klimek, Greenberg, Eleff, & Thompson, 1992) and worry about children's risk (Dudok de Wit, 1997), which may not register as significant on these questionnaires, highlighting the contribution of a more qualitative approach.

The research literature can be confusing when attempting to identify family members at risk for

adverse reactions to PGT in a clinical setting, and there is a lack of theoretical models specific to high risk families (Cull, Anderson, Campbell, Mackay, Smyth, & Steel, 1999; McAllister, 2001; Rees, Fry, & Cull, 2001; Reeve, Owens, & Winship, 2000). Traditional health behaviour models (e.g. Becker, 1974; Leventhal, Benyamini, & Brownlee, 1997; Leventhal, Mayer, & Nerenz, 1980; Rosenstock, 1974) have been used in attempts to predict screening behaviour. However, findings to date are somewhat contradictory (Conner & Norman, 1996), and it is difficult to use any of these models to predict what high risk family members might do with regard to PGT. In addition, many of these models have been developed with the general aim of predicting and improving compliance with health-promoting behaviour, and may not be appropriate for the specific case of PGT in high risk families, which, for most conditions, does not promote health in any direct way. However, it could be argued that people knowing they are mutation carriers may have an impact on screening behaviour in high risk cancer families.

It is important both theoretically, and for development of clinical services in genetics to develop social psychological models that have explanatory power (McAllister 2001; Michie & Marteau, 1996; Rees et al., 2001). This research was an attempt to contribute towards the filling of this gap. Since the severity of the particular condition can influence behaviour around PGT (Evans et al., 1997), it was decided to focus on one condition only for which testing is available.

Hereditary Non-Polyposis Colorectal Cancer (HNPCC) is a dominantly inherited cancer syndrome, predisposing mutation carriers to a high risk of early-onset colorectal cancer in both sexes (74 per cent), endometrial cancer in women (42 per cent), as well as increased (but lower) risks for other cancers such as ovarian, gastric, urologic tract, small bowel, hepatobiliary tract, and brain tumours (Watson & Lynch, 2001). The risk for colorectal cancer and perhaps also for endometrial cancer, can be reduced by regular screening, but the screening procedures are invasive, uncomfortable, and carry risks of their own. Possible motives for PGT in HNPCC families include: (1) the wish to clarify the position for children; and (2) to avoid unnecessary screening. Motives against PGT might include perceived

incapacity to live with the knowledge of such a high risk for cancer.

The broad research question addressed in the present study was: What social psychological process explains variations in HNPCC family members' approaches and reactions to PGT? The specific aim in this study was to build social psychological theory that would have some explanatory value with regard to variations in adjustment to PGT. The research began with a broad base, with one open directive research question, and some ideas about what kinds of issues *might* emerge; there were no initial hypotheses (McAllister, 1999).

Methodology

Strategy

The methodology used in the present study was grounded theory approach (Glaser & Strauss, 1967; Strauss & Corbin, 1998), which is a means of building social psychological theory using the data of qualitative research. The way in which grounded theory was used in this study is described in detail in McAllister (2001). Since the aim in this research was to build theory, rather than merely to describe themes in the data, grounded theory was chosen over other qualitative research methods. For the most part, attempts were made to maintain a realist analytic approach to the study; however, a degree of interpretation was unavoidable. This was particularly the case where the analyst interpreted the social function of the interviews in creating identities for the participants. At such points, a more discursive interpretation was made. In addition, a knowledge of the literature, and theoretical writing about families and genetic disease (e.g. Richards, 1996) was used to inform some of the interpretations made.

Recruitment and interviews

Approval was obtained from local medical research ethics committees and a national psychological research ethics committee prior to recruitment. Patients were identified and recruited by the clinical team. Recruitment involved sending an invitation letter, information sheet, and consent form to these patients. Consenting patients were then contacted directly, and a time for interview arranged. Twelve families were recruited through three hospital genetics

departments and one hospital gastroenterology department, and later family 'snowballing' (see Table 1). In nine families, the underlying

Table 1. Sample characteristics
N = 55 (Total number of families = 12)

Male	25 (45.5%)
Female	30 (54.5%)
Married	46 (83.6%)
Single	4 (7.3%)
Divorced	5 (9.1%)
No children	9 (16.4%)
Adult children	29 (52.7%)
Young children	17 (30.9%)
Affected with cancer	9 (16.5%)
At risk	15 (27.3%)
Unaffected carriers	7 (12.7%)
Non-carriers	8 (14.9%)
Married in (not at risk)	16 (29.1%)
Age 18-29	3 (5.5%)
Age 30-39	12 (21.8%)
Age 40-49	12 (21.8%)
Age 50-59	17 (30.9%)
Age 60-69	9 (16.5%)
Age 70-79	2 (3.6%)

(McAllister, 1999)

HNPCC mutation had been identified and PGT could be offered, and in the other three families a mutation search was underway, but had not yet been successful, so PGT was not (yet) available. Fifty five family members were interviewed, including 12 individuals who were followed through PGT (see Table 2). Interviews took place in participants' homes. Participants were interviewed alone in all cases but two (one where a sibling came into the room a few minutes before the end, and interjected; and another, where a husband and wife insisted upon being interviewed together). The interviews were recorded (with consent) and took on average 45 minutes (range 20-110 minutes). An 'interview guide' was used (see Appendix 1).

Data analysis

Interviews were transcribed and analysed using grounded theory approach, supported by ATLASTi qualitative analysis software (Scolari Scientific Software Development, 1997). See McAllister (2001) for elaboration of the tools of grounded theory and their application in this study.

Table 2. The interviews

<i>Predictive testing families: 8 families where the underlying mutation had been identified and family members could have predictive testing</i>	<i>Interviews</i>	<i>Mutation search families: 3 families where the underlying mutation had not yet been found so family members could not have predictive testing</i>	<i>Interviews</i>
12 individuals from 7 families followed through predictive testing	38	12 family members interviewed once only	12
<ul style="list-style-type: none"> • Interview 1: prior to test result (but after genetic counselling) • Interview 2: 1–2 weeks after test result • Interview 3: 6 months after test result 		<ul style="list-style-type: none"> • 2 family members affected with cancer • 9 at risk family members • 1 spouse 	
2/12 interviewed again 12 months later			
19 individuals from those 7 families NOT being tested (spouses, adult children and declining siblings) interviewed once only	19		
10 individuals from an 8th family where predictive testing had been completed interviewed once only	10		
2 individuals from a 9th family interviewed prior to predictive testing but later withdrew from the study	2		
Sub-totals	67		12

Total number of interviews = 81

Results

The emergent theory in the present study is concerned with: (1) explaining how the postulated process of engaging with HNPCC risk is proposed to take place; and (2) how the degree of engagement that has occurred at the time of PGT is proposed to influence approaches and reactions to test results. In this paper, the theory will be presented in overview.

A core category of *engagement* (see below) emerged in the present study as having a significant influence on approaches and reactions to PGT for HNPCC (see McAllister, 2001). The resulting theory is built around the concept of *engagement*, and the associated process of *engaging with HNPCC risk* (see Fig. 1). Engagement is a new theoretical construct, which may have explanatory value with regard to variations in attitude towards one's risk status. The degree of engagement may change with the unfolding of time and HNPCC-related events in family life.

The process of engaging with HNPCC risk is proposed to occur in HNPCC family members over time, as they become cognitively and emotionally involved with their risk of cancer as a result of their interpretation of their family history of cancer. As the process of engaging with HNPCC risk occurs, the degree of engagement may change, and the attitude towards one's risk status and PGT may change. At any given point in time, an individual in a HNPCC family may either be completely unengaged (ie. they have not yet begun to engage with their risk of cancer), or they may have engaged to some degree along a continuum from very partial engagement to very intense engagement (see Fig. 2).

The concept of engagement

Engagement is an interpretive code (Strauss, 1989). The conceptual label and definition of 'engagement' were devised by the researcher as the most appropriate descriptors of the data

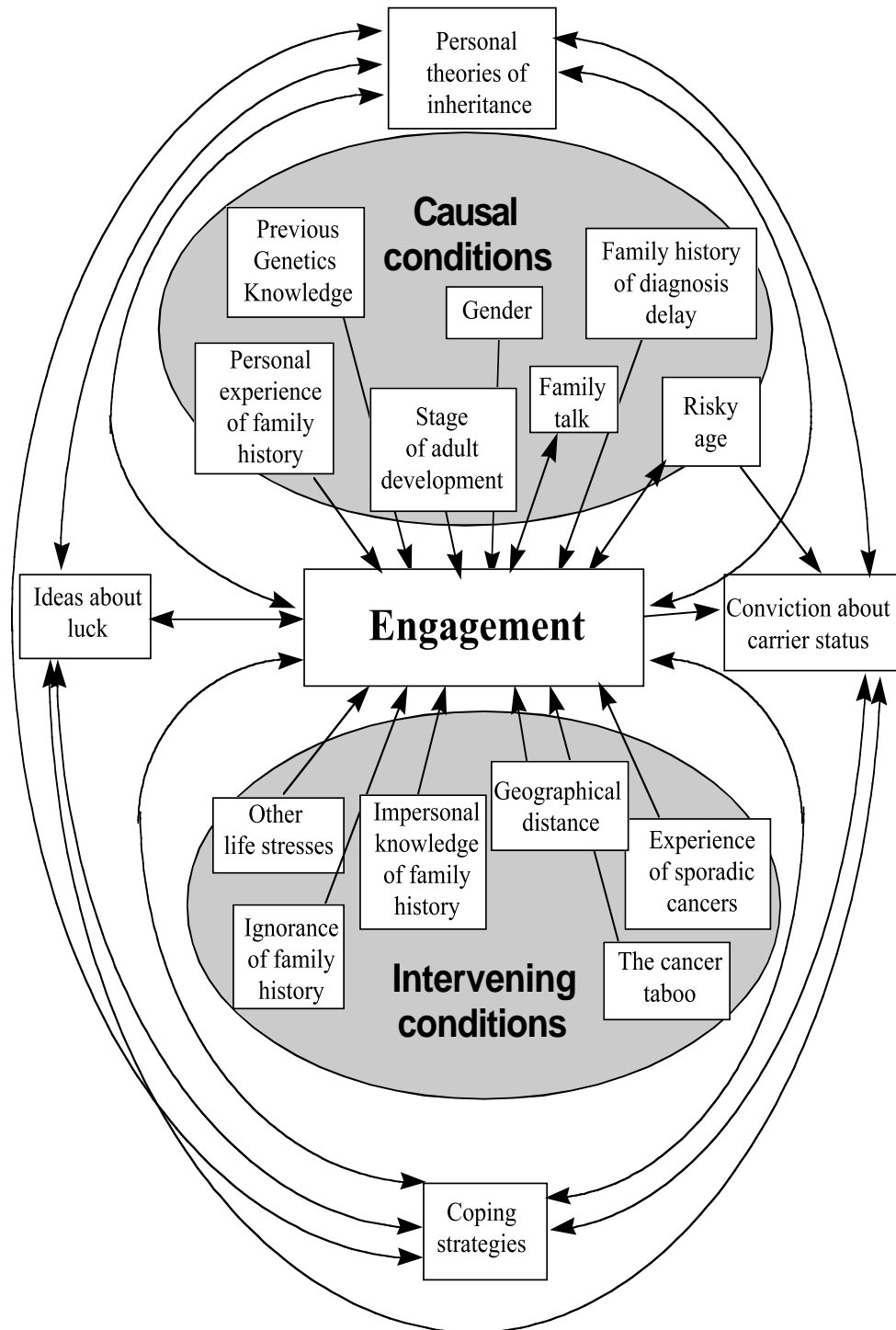


Figure 1. Process of engaging with HNPCC risk (McAllister, 1999; McAllister, 2001).

through an evolving process of data analysis, beginning with open coding, and developing through theoretical sampling, constant comparison, axial coding and selective coding. The way in which the definition of concepts in this study was developed is described in detail in McAllister (2001). Engagement was defined as the degree of cognitive and emotional involvement with one's increased risk of developing cancer as a result of one's family history of cancer and the concept was confirmed and developed using the principles of grounded theory as the study progressed. The emerging theory of engagement (TE) postulates that engagement influences attitudes towards PGT. Individuals in HNPCC families may be engaged at the cognitive level only. In the present study, these individuals (see Q1) did not express fear or anxiety in relation to their risk status, and are referred to as *partially engaged*.

Q1: I don't suppose it'd sink in . . . unless I had cancer . . . you know . . . because, to me, at the moment, it's just like, 'Oh, I'll just get tested . . .' As far as my brother goes . . . emmm . . . I don't know . . . it didn't really—I don't want to sound nasty—bother me.

Other individuals (see Q2) in the study expressed some fear/anxiety in relation to their risk status; they are described as engaged at the cognitive and affective levels and are referred to as *intensely engaged*.

Q2: I was imagining things the last couple of months—every time I got a stomach-ache, or—you think 'Oooh, I wonder if something . . . you know'—you do—you definitely do—when you're at risk of something—you tend to make something out of nothing really, if you're feeling a bit under the weather . . .

Engagement at the affective level only was not seen in the present study; cognitive engagement is suggested to be a necessary but insufficient precursor to affective engagement. This is consistent with established thinking on cognition and affect (Lazarus, 1982, 1991; Power & Dalglish, 1997). The degree of engagement in the present study had a consistent association with action in relation to cancer risk, and this is illustrated in Fig. 2.

As shown in Fig. 2, there is a theoretical continuum from unengagement (engagement has

not yet occurred) through partial engagement to intense engagement. The TE postulates that no action is taken in relation to cancer risk without some degree of engagement. Furthermore, a critical degree of engagement (not defined in this study) may be required before any action will be taken. By the time a family member presents at a genetics clinic, it is assumed that some engagement has already occurred, because presumably one would know why one was attending, and would have thought about this, even if only superficially. As this paper will demonstrate, the *degree* of engagement at a given time may help explain how an individual in a HNPCC family will approach and react to PGT.

Disengagement describes the observation in the present study that individuals in HNPCC families who show signs of having been intensely engaged can cut themselves off from thoughts, feelings and action in relation to their risk. Similar to avoidance/denial, disengagement appears to occur when engagement involves the experience of intensely painful emotions. Because of this, engagement may be avoided (Q3).

Q3: I mean, I guess on one level I sort of knew . . . partly, I wonder whether I'd sort of subconsciously . . . not—forced myself not to look at it [. . .] because I tend to be the sort of person who just—if something wasn't . . . I just blot it out.

Disengagement may be reversible. Disengaged family members may become engaged again in the future; they may at times oscillate between intense engagement and disengagement. This kind of oscillation can occur in individuals who are attempting to come to terms with something which they have been denying (Maguire and Faulkner, 1988).

Process of engaging with HNPCC risk

Engaging with HNPCC risk (see Fig. 2) may be a difficult process for family members as it involves dealing with painful memories, e.g. death of a parent at a vulnerable age. Because of this, progression of engagement with HNPCC risk may be avoided, or it may be a process that occurs slowly over the lifecourse. A critical event (see Q4) may be required to precipitate or

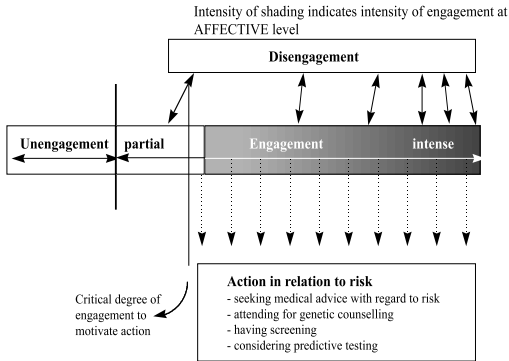


Figure 2. Engagement and action in relation to risk (McAllister, 1999).

progress the process, for example a new diagnosis of cancer in the family.

Q4: I suppose I realised there was something a bit strange simply from the history of (my husband’s) sister and her daughter who died—and then when the other daughter developed ovarian carcinoma, (my husband) and I discussed it, and I said ‘well, there’s obviously a gene in that side of the family’.

It is suggested here that the degree of engagement in an individual HNPCC family member may change with the unfolding of events over time, or on further reflection over time about their situation or about the family history (Q5): this is the postulated process of *engaging with HNPCC risk* (see Fig 2).

Q5: I suppose that was the explanation I would give for not having got on quicker with having the (colonoscopies) done . . . at that stage—I wasn’t as clued up, or as informed about it . . . and then there is what’s since happened (*bowel perforated on routine colonoscopy, and as a result, a large part of bowel surgically removed*) it was something, then, that wasn’t of great importance. It’s increased in importance since then. I didn’t address the thing, then, as much as I ought to have done [. . .] I hadn’t properly appreciated the importance of it.

Interviewer: Why do you think you are able to appreciate that now?

Oh, because I’ve thought about it a lot more . . .

Engaging with HNPCC risk is postulated to be a dynamic process that occurs initially at the cognitive level (partial engagement). With the passage of time and events, engagement may progress at the affective level leading to intense engagement (and sometimes disengagement), as the man quoted above (Q5) said in a later interview (Q6).

Q6: . . . it wasn’t something that was at the top of my mind the whole time while I was waiting for the result . . . it was only when I knew that they had the results that I started to get anxious about what the results were. [. . .] I was apprehensive about what the results were, and how things were going to work out.

A series of psychosocial factors were identified in the present study as influencing this suggested process of engaging with HNPCC risk. These are illustrated in Fig. 1, and include: (1) causal conditions which facilitate the process such as personal experience of, and family talk about the family history of cancer; (2) intervening conditions which block the process such as ignorance of the family history, other life stresses and experience of sporadic cancers; and (3) individual psychological factors such as personal theories of inheritance and coping strategies. The individual factors that may influence the process of engaging with HNPCC risk are described in detail elsewhere (McAllister, 1999). At this point, there are three important points to note.

1. *The postulated process is a psychosocial one.* For example, as illustrated by Q7, when the family does not discuss the family history, family members can forget about it and remain partially engaged. In contrast, Q8 shows a quote from a lady who was very frightened about her risk of developing cancer in later life, and describes how her mother talked about the family history in her youth.

Q7: . . . time sort of moves on . . . and it just sort of drops to the back of your mind when there’s no-one going on about it. You just hope that it will go away really (*partially engaged*).

Q8: The first I heard of it was [. . .] when I was

at home, as a girl, you know . . . because—(my mother) lost her father with the cancer of the bowel [. . .] He died—I think he was 30 [. . .] and what a terrible ordeal—you know—what a terrible thing it was in them days [. . .] that's the things I heard about from me mother . . . (*intensely engaged*)

An interaction is suggested between gender and family talk, and between gender and engagement status, in that women tend to talk about family history more than men do (see also McAllister, Evans, Ormiston, & Daly, 1998), as illustrated by quotations Q9 and Q10 from a husband and wife.

Q9: (my at risk husband) doesn't really go much into family history—even now, he couldn't tell you who his cousins are . . . or his uncles. . . . But I'm quite interested in that. . . . Any information that I have got would be from speaking to his mother on the phone, or his sisters. I mean, I'm not just talking about cancer, I'm talking about divorces! (Laughs) Everything! [. . .] So, I mean, he would discuss it—but superficially. I think that once feelings got in the way, it would pretty much be—end of the discussion (*intensely engaged*).

Her husband said:

Q10: (my wife) will know more about that than me because she takes . . . she's interested in things like this . . . and she'll chat to my mother about the family . . . whereas . . . we just don't bother asking the obvious questions. So that might be something maybe to follow up with her [. . .] as I say I'm pretty clueless when it comes to family—(my wife) knows better than me what's what in the family (*partially engaged*).

It may be that some participants were, at times, using the interviews to create gendered identities for themselves, such as the feminine 'genetic housekeeper' (Richards, 1996). Issues of gender may, therefore, *interact* with engagement status, rather than influencing it directly (see Fig. 1).

Some family members in the present study expressed 'personal theories of inheritance', which predicted for them who in the family will inherit the gene. These theories appeared to be strongly held only by family members who were intensely engaged (see Q11), and who, (in the

present study), all believed themselves to carry the family mutation. Such theories appeared to: (1) be specific to *individuals*, rather than *shared* in families; and (2) draw upon existing lay models of inheritance based mainly upon co-inheritance of cancer (predisposition) with some other physical characteristic in the family. Although partially engaged family members may also hold such theories, in the present study they were held less firmly, and these family members did not have strong convictions about their own or other family members' carrier status. Partially engaged family members appeared to find it easier to accept a 50 percent risk (Q12).

Q11: (the genetic counsellor) explained it as being two sets of—is it chromosomes?—one good one, one bad one—and you're going to inherit either the good one or the bad one—emmm . . . either the good one from your father or the bad one from your mother. Sooo . . . I understand it that way. You've got a 50-50 chance of having the good one or the bad one. But we tend to think the bad one's more—sort of—dominant over the other one—we all think—we're all sure that we're all going to get it (*intensely engaged*).

Q12: It's just 50/50 chance—one could have it, one might not, or we might both have it, or neither of us—just a chance either way, really (*partially engaged*).

The interpretation here is that there may be a dynamic interaction between personal theories of inheritance and engagement status (see Fig. 1).

2. *Many of the social factors influencing engagement seem to be related to the experiences of family life.*

The personal experience that family members have of the disease in their family may influence engagement status. The outcome of cancer in close family members—personal experience of cancer suffering—appeared to facilitate progression of engagement in the present study. Intense engagement tended to be seen where close family members had been witnessed suffering or dying from cancer. If relatives had survived cancer without too much suffering, then engagement tended to remain partial only (see

Q13). However, in a couple of cases where the emotional experience was particularly intense, for example where a close family member was witnessed dying from cancer at a vulnerable age, disengagement was seen (see Q14).

Q13: . . . illness—you just take in your stride—God's good—He's got (my brother) through it—He's got (my husband) through it—[. . .] got me mum through it—it was smoking that did it in the end—as opposed to the genetic cancer—you know—I mean, they worked miracles with that colostomy—they did . . . (*partially engaged*).

Q14: . . . he was doing this ultrasound scan and he said 'emmmm . . . have you had any children?' and I said 'no, I'm not married' and I'm just lying there, you know . . . eh . . . and he said 'oh, well your womb's very large' and I said 'well, I haven't got one' because I'd had a hysterectomy . . . because I'd had very bad period trouble, so I'd had that oh . . . 5 years prior to that . . . and he said 'Oh . . . well there seems to be a large growth or something here . . .' Of course, immediately, like immediately, all the pennies sort of dropped . . . because my sister's cancer had been ovarian . . . that's when I knew it was hereditary (*previously disengaged; sister died in her 20s; mother also affected*).

3. *The psychological factors are postulated to interact with each other and with the social factors in the model as the process of engaging with HNPCC risk progresses.*

Harre & Secord (1972) have argued that accounts are not reflections of causes *per se*, but are *post hoc* justifications for actions. Perhaps, similarly, with regard to beliefs about their mutation carrier status rather than actions, HNPCC family members cite 'explanations' for their beliefs, which are actually justifications for their 'preferred' coping strategy (see also Richards, 1996). Quotations Q15 and Q16 were interpreted as individuals using such explanations to support a preferred coping strategy.

Q15: I had taken it on board that . . . I probably did have the gene. I think that was a way of dealing with things for me. Rather than having the uncertainty of thinking 'have I got it? Have I not got it?', it seemed a lot better just to say 'I've probably got it' and get on with

life, and cope with that and deal with it . . . That's my way of dealing with things—prepare for the worst—hope for the best (*intensely engaged*).

Q16: . . . it's a good 50–50 chance. But . . . I'm very much like my father in all respects—so that's the positive side for me—(my eldest sister) is more like my mum—I tend to be more like the (*foreign*) side—so I think there—that's the positive side of it.

Interviewer: You think that will go in your favour?

I think it will do. Yes—I think it will do—I really do. . . . I'm very much like my father's side. (My youngest sister)—is half and half—but I know myself that I'm more like the—you know—the (*foreign*) side—as happy as the day is long (*partially engaged*).

Beliefs about luck (Q17) can also come into play.

Q17: I [. . .] think I will have the faulty gene. Yeah—just my luck—(laughs)—I'm just an unlucky person (*intensely engaged*).

To put this in a slightly different way, perhaps HNPCC family members develop narratives around their risk status that enable them to cope in a way which fits their 'preferred' coping strategy. The model suggested here is that personal theories of inheritance, ideas about luck, coping strategies, and conviction about carrier status all interact *recursively* with each other, and with degree of engagement to influence risk perception in HNPCC family members having PGT (see Fig. 1).

Engagement status is proposed to develop and to be maintained in interaction with a 'preferred' coping strategy involving increasing attention to, and reflection about certain aspects of the family history of cancer and cancer risk. Where partial engagement progresses towards intense engagement, attention may be selectively directed at the negative implications of the family history, perhaps leading to increasing cycles of cognition resulting in the generation of anxiety (Tallis & Eysenck, cited in Eysenck, 1992). It is proposed that this process may be facilitated by interaction with ideas about luck, and selection from a set of existing schemas about biological inheritance, one that predicts

mutation carrier status, and maintenance of this as the dominant schematic model. This is consistent with work on information-processing in anxious individuals which shows biases in favour of threat-related material (Mogg, Mathews, & Weinman, 1989; Mogg, Mathews, Eysenck, & May, 1991a), and increased accessibility amongst anxious people of reasons why a negative event would happen to them, relative to accessibility of reasons why it would not (MacLeod, Williams, & Bekerian, 1991). Other studies have shown that anxiety increases subjective risk estimates (Johnson & Tversky, 1983). Rees et al. (2001) used a similar argument to predict theoretically that women's personal experience of breast cancer in their family can influence their risk perception. In this way, for intensely engaged family members, a coping strategy of 'preparing for the worst' may influence risk perception by facilitating generation of a narrative of the self as a mutation carrier.

Inhibition of alternative schematic models or information have been included in recent cognitive theories of emotion, such as SPAARS (Schematic, Propositional, Analogical, and Associative Representation Systems) (Power & Dalgiesh, 1997) as a means of maintaining cognitive and emotional order. This may explain the apparent (conscious or unconscious) 'rejection' of the mendelian explanation of inheritance amongst the intensely engaged. Perhaps, the genetic model does not 'fit' with their 'preferred' coping strategy of working through coming to terms with mutation carrier status prior to obtaining their PGT results; yet they experience a need to maintain internal cognitive consistency. Perhaps they find some other explanation for inheritance that enables them to cope in their preferred way, whilst at the same time, maintaining internal cognitive consistency. In generating this narrative, it is suggested here that family members draw upon existing schemas about how biological inheritance operates. Intensely engaged family members in the present study seemed to use a variety of models or schemas (e.g. a sex-linked schema, or perhaps one based on co-inheritance of cancer predisposition with some other physical feature), to predict their own carrier status in a way which seemed to best fit their own preferred coping strategy.

The suggestion made here is that for the most

part, the process of engaging with HNPCC risk may occur in the direction of more intense engagement over time as HNPCC family members become increasingly aware of their own risk. As HNPCC-associated events unfold in family life, individuals may first become partially engaged (engaged at the cognitive level only). They may then proceed to become engaged at the affective level (intensely engaged). If intense engagement is experienced as too painful, disengagement may occur. However, it is also suggested that the intensity of engagement may decrease with time. This may be related to aging, particularly when family members safely pass the age at which their close relatives were affected (see Q18). Family members may thus move from partial engagement to intense engagement over time, and then back to partial engagement.

Q18: Sometimes it used to play on my mind . . . I'd think . . . my mother died when she was 54 . . . my sister says 'she wasn't—she was 58', but she wasn't—she was 54—I know. And now—I've got past me mother's age . . . so I feel all right now.

Consequences of engagement status for adjustment to predictive testing

The degree of engagement prior to testing emerged in this study as associating consistently with approaches and responses to PGT for HNPCC (see Tables 3 and 4). Because the study was longitudinal, engagement status had been designated by the analyst prior to test result.

Approaches to testing

In the present study, whether participants believed themselves to be mutation carriers because of the predictions of a personal theory, or because it fit with a preferred coping strategy, the consequences for intensely engaged family members appeared to be the same: they seemed to think and feel as if they were mutation carriers—they appeared to be 'rehearsing' mutation carrier status, or to be doing 'the work of worry' (Binedell, Soldan, & Harper, 1998a,b; Dudok de Wit, 1997; Janis, 1958). This was interpreted as reflecting that they had come to terms with mutation carrier status *before* they obtained their test result (see Q19 and Table 3).

Q19: It was concerning . . . But by this time it was my belief that if you're getting the medical attention . . . it's always been there—we've not changed anything, so . . . we've got the medical contacts now . . . we could have buried our head again . . . this has happened; I'm the type of person who wants to do as much as I can to resolve things . . . so all the way through, I was being positive towards (doctor) and the letters . . .

hand, did not seem to have strong beliefs about their carrier status; they appeared to accept the 50 percent risk, and they did not seem to do 'the work of worry' prior to obtaining their test result. Intensely engaged family members appeared to view the test as a gateway to screening for a form of cancer, which is common in the general population, even before they obtained their test results. The emerging hypothesis is that those who are intensely engaged have 'rehearsed' mutation carrier status; they have worked through the implications and have come

Partially engaged family members, on the other

Table 3. Consequences of engagement: proposed relationships between engagement status and approaches and reactions to HNPCC predictive test results

	<i>Partial engagement</i>	<i>Intense engagement</i>	<i>Disengagement</i>
<i>Definition</i>	Thoughts about FH cancer. Engagement at the cognitive level only.	Thoughts and feelings about FH cancer. Engagement at cognitive and affective levels.	Avoid thoughts and feelings about FH cancer. No CURRENT engagement at either cognitive or affective levels.
<i>Prior to test result</i>	Do not experience anxiety/fear. Adopt wait and see approach to test result. Accept 50/50 risk.	Experience anxiety / fear; expect bad result. Come to terms with mutation carrier status prior to test result.	May oscillate with intense engagement. Do not present for predictive testing whilst disengaged, although oscillation may be seen during the testing protocol.
<i>1-2 weeks after GOOD test result</i>	Rather blasé. Relief in relation to childrens' health.	Disbelief. Intense relief in relation to their own and their childrens' health.	N/A
<i>6 months after GOOD test result</i>	No difference—all have come to terms with test result IF carrier siblings are still healthy.		N/A
<i>1-2 weeks after BAD test result</i>	Experience anxiety.	Accept test result quickly and easily.	N/A
<i>6 months after BAD test result</i>	No difference—all have come to terms with test result IF clear screen.		N/A

Table 4. Consequences of engagement: proposed relationships between prior engagement status and reactions to HNPCC predictive test results

<i>Prior engagement status</i>	<i>Predictive genetic test result</i>	<i>1-2 week post-test adjustment</i>
Partial	Mutation positive	Poor
Intense	Mutation positive	Good
Partial	Mutation negative	Good
Intense	Mutation negative	Fair

to see carrier status in a positive light (see Q20 and Table 3).

Q20: Myself and my sister think that we're all likely to have it. So—it won't be a big shock if we have—but it would be nice to know—so that we can have the screening or whatever—cos [. . .] it is treatable.

Reactions to test results

(1) *Prior intense engagement:*

Those family members in the present study who were intensely engaged prior to obtaining their PGT result *and* were found to carry the mutation, such as the man quoted at Q21 below, appeared to accept their carrier status quickly and easily.

Q21: (*doctor*) unfortunately said that I have inherited my mum's gene. But again—there was no tears, or sadness, or anything like that—it just confirmed to me what I have thought for the last 9 months [. . .] technically, whilst I've got greater odds of getting it, I've got a greater percentage chance of surviving it because it will be found in me before it develops, whereas the guy on the street will only find out once it has developed into a more advanced stage.

These individuals seemed to have developed a positive attitude towards mutation carrier status prior to obtaining their test result. Those who were found *not* to carry the family HNPCC mutation, such as the woman quoted at Q22 below, appeared to experience some disbelief as well as intense relief in the initial post-test period.

Q22: I really thought I'd be positive—I got more of a shock being negative than I would have done being positive.

The interpretation made here is that the anxiety and worry experienced by the intensely engaged prior to obtaining their test results may be *functional*, at least for those who are found to carry the mutation (see Table 3).

(2) *Prior partial engagement:*

In contrast, family members with prior partial engagement who were found to carry the family HNPCC mutation appeared to experience some anxiety in the initial post-test period (see Q23). This anxiety, although seeming rather

non-specific, can be considered sufficiently intense to require medical attention.

Q23: I did feel sort of a bit funny over the weekend—I felt a bit sort of anxious . . . but I don't know whether it was because of that or because of other things [. . .] I made an appointment to see the doctor, but he was booked up, and by the time I saw him, I felt a bit better [. . .] I suppose I'll have to have the colonoscopy now—I'm not relishing that.

Non-carriers with prior partial engagement seemed to accept their test results quickly and easily (see Q24), appearing sometimes rather blasé about it at two weeks post-testing.

Q24: . . . I was glad because of the kids. I don't have to worry about them. It hasn't been passed down to them. That's all. I didn't feel much else—I wasn't that bothered. . . . I didn't want to go through all them tests—they're not very nice.

However, six months after PGT, all those tested in this study seemed to have come to share the view that mutation carrier status is a gateway to screening for a common form of cancer, regardless of prior engagement status. Adjustment seemed to be related to the experience of screening, as illustrated by Q25 and Q26.

Q25: *Prior intense engagement:* I just think we're lucky we had that blood test. I mean—even if there is something there, it wouldn't have been found otherwise, would it? So, I mean, I think we've been lucky—very lucky.

Q26: *Prior partial engagement:* So—I've got a gene that's inherited and . . . but they were able to tell me that I'm OK inside—which is great to know. Because it's that warning, now, to you. You've been warned in advance—and it's up to us and the hospital to keep an eye and a check. It's marvellous—my mum never had the opportunity but we have.

Similarly, amongst those found *not* to carry the family mutation, differences were no longer apparent at six months post-testing (see Table 3). Like other workers (eg. Michie, McDonald, & Marteau, 1996), this study (Q27) showed that family members may regret the loss of screening:

Q27: As the day (*that I got my result*) went on, I got higher and higher and higher . . . but

there was this sort of feeling of anticlimax as well . . . it was finished with—over with—no more tests. Then the fear set in—well—you know—what if I'm one of those other people that gets it without having the gene? Because what they said was if you haven't got the gene, then you've got the same chance as anybody else in the population now of having it—so maybe I'm one of those people now. . . .

In the two cases of this in the present study, the individuals had never had a colonoscopy. Their feelings about losing the opportunity to have colonoscopies could not be explained by habituation to regular reassurance obtained from prior colonoscopies, as in the Michie et al. (1996) study. There was also no evidence that the test result had failed to alter their perceived risk of developing cancer, another possible explanation offered by Michie et al. (1996) to explain the FAP findings. A possible explanation for the reaction of the two women in the present study emerged from the data. The two who felt anxious because they were losing their screening as a result of their non-carrier status both had siblings for whom HNPCC carrier status was currently an issue.

Q28: It didn't enter my mind to start with . . . it was only afterwards I thought . . . cos (*my carrier sister*) was talking about arranging to have her screening done . . . and I thought 'Oh, bloody hell, I won't be getting that now'.

However, numbers are very small here, and so interpretation must be very tentative.

Discussion

This was a qualitative study aimed at building theory that might provide some explanation of behaviour around PGT for HNPCC. Although the sample size was small, and only 12 individuals were followed through PGT, some consistent findings emerged enabling a very tentative theory to be developed. However, the small sample size, particularly in the longitudinal part of the study, is a major limitation, and more research is needed to further develop and confirm (or refute) the TE. At present, the results from this study cannot be generalized to other populations. It should also be mentioned that alternative interpretations of the interviews

might have been made by others with alternative analytic positions.

The plausibility of the TE is supported by much previous research. Many of the phenomena that emerged as potentially influencing attitudes towards PGT for HNPCC in the present study, such as family experience of the disease, higher perceived risk and lay beliefs about how the disease is inherited in the family, were not altogether unexpected (Dudok de Wit, 1997; Geller, Doksum, Bernhardt, & Metz, 1999; Rees et al., 2001; Richards, 1996; Rolland, 1994). The present study lends further support to this work. However, the TE provides a possible explanation of *how* these factors interact, through the process of engaging with HNPCC risk, to influence attitudes towards PGT. In addition, the TE introduces an explanation of change over time through the postulated process of engaging with HNPCC risk.

The findings in the present study are also consistent with previous findings by Reeve et al. (2000) and Michie et al. (1996), with families having PGT for inherited colorectal cancer. These studies: (a) did not suggest a simple model whereby a good psychological outcome could be expected for those found not to carry the gene mutation, and a poor psychological outcome for those found to be mutation carriers; and (b) indicated that a focus on colonoscopy was important as a way of coming to terms with mutation carrier status. Furthermore, the present study extends this work by presenting a possible explanatory model that may help practitioners to understand why some family members find it easier to come to terms with a 'bad' test result than others, and provides some potential indicators to help identify those who may be at risk for a short-term adverse reaction following test results.

It has been suggested previously that distress prior to PGT might be functional, in that it might reflect a working through of the possible implications of a 'bad' result (Binedell et al., 1998a, b; Dudok de Wit, 1997). In the present study, prior intense engagement is suggested to be associated with ease of adjustment to a bad test result, providing support for this idea, and developing it by offering a plausible explanation for *how*, through the proposed process of engaging with HNPCC risk, intensely engaged HNPCC family members appear to come to terms with mutation carrier status prior to PGT.

Some elements of the TE are supported by recent research studies on the psychosocial impact of PGT, although, unfortunately, there are no suitable comparable studies with HNPCC families. Studies to date with HNPCC families have been small, and have not identified any useful correlations between pre-test predictors and post-test distress (e.g. Lynch, Lemon, Karr, et al., 1997; Reeve et al., 2000). With regard to research on other inherited disorders, which may differ from HNPCC, the influence of approaching the affected parent's age at onset in increasing risk perception was shown by Decruyenaere, Evers-Kiebooms, Boogaerts, Cassiman, Cloostermans, Demyttenaere, Dom, & Fryns (1999) in a study of HD family members offered PGT. A similar relationship is suggested in the present study, in that approaching the affected parent's age at onset may facilitate progression towards intense engagement, with an associated increase in risk perception.

However, there are important elements of the TE that are not supported by the results of other studies, particularly the inverse relationship identified in the present study between pre-test engagement status, test results, and post-test distress. Some studies with HD and other families have shown that pre-test anxiety is the best predictor of post-test distress, irrespective of the test result (eg. Decruyenaere, et al., 1999; Dudok de Wit, 1997). In contrast, the TE predicts (1) that individuals who are intensely engaged (ie. who experience fear and anxiety, and who believe themselves to carry the family mutation) prior to testing do well following testing, even if the test result is unfavourable; and (2) that individuals who are partially engaged (do not experience fear and anxiety, and do not have strong beliefs about carrier status) prior to testing *do* experience distress following testing, but only if they obtain a bad result. Previous studies looking at pre-test predictors of post-test distress for a number of different disorders have been somewhat inconclusive (Broadstock et al., 2000), and this *may* be because the pre-test psychological measures used were too general for the specific context of predictive testing for cancer.

The TE was built using the experiences of families with a high risk of developing cancer, and includes concepts not taken into account in

many of the previous studies, such as the concept of engagement, the degree of personal experience of the family history of cancer, and family communication. Although the present study suffers from a major limitation in terms of sample size, because the tentative theory that emerged is: (1) grounded in the real-world experience of HNPCC families having predictive genetic testing; and (2) provides a plausible explanation for variation in behaviour around predictive genetic testing, further research would seem to be justified in an attempt to either confirm, refute or revise the concepts and inter-relationships postulated in the present study.

To this end, a further study is planned to develop and validate a multi-item scale to test the theory in relation to predictive testing, in a large quantitative study. It is hoped that this study will provide data to further develop and refine the TE. The theory of engagement may potentially have wider implications for research, and could benefit from further development with other inherited disorders such as hereditary breast/ovarian cancer and Huntington's disease. The 'engagement with familial risk' scale may prove to be a useful research tool in these contexts, for exploring if and how the dynamics of engagement operate in these and other inherited conditions. The TE indicates that reactions to predictive test results are focused on the benefits of screening, and we know that uptake of testing is higher for conditions where treatment is available (Evans et al., 1997). In addition, it has been suggested that potential availability of treatment may diminish concerns about passing on the risk of HD (Downing, 2001). With further data collection and theoretical refinement, the TE might provide a plausible integrated theoretical explanation for this variation. The theory would be likely to be substantially altered with increasing development (Glaser & Strauss, 1967).

In conclusion, although the small sample size in this study was a major limitation, the emerging TE provides the first evidence-based psychosocial process model explaining variations in behaviour around PGT for cancer. The findings provide a useful framework for further research, as well as potential implications for clinical practice in genetics.

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Appendix

Interview Guide (McAllister, 1999)

HNPCC: A Family Study
Interview Guide

Ask them to tell their own story:

Can you tell me in your own words the story of what has happened in the family as regards cancer?

- Who in your family has had cancer?
- How well did you know them?
- Did you see much of them while they were ill?

Interviewer should be able to sketch a family tree.

Then elaborate on themes:

1. *Inheritance beliefs, e.g.*

What do you think causes HNPCC?

Can you describe to me how the faulty gene is inherited?

Do you have any gut feelings about whether you or anyone else in your family is more likely to have the faulty gene than anyone else?

Why do you think that?

2. *Genetic counselling, e.g.*

Are you (or is anyone in your family) having any genetic counselling?

How did that come about?

What did you learn?

3. *Screening, e.g.*

Are you having any screening?

(If not) What are your reasons for not wanting to have screening?

What about other people in your family?

How do you feel about screening?

4. *Testing, e.g.*

How did you first find out about the predictive genetic test?

Has anyone in your family had a predictive test?

How do you feel about testing?

Is this something that you are considering?

(If so) What are your reasons for wanting to have a test?

(If not) What are your reasons for not wanting to have a test?

Following test results:

Can you remember your first reaction when you found out your result?

How do you feel about it now?

Are you glad that you had the test?

Have your feelings about your brothers and sisters changed in any way since the results of testing? Parents? Children?

What has been the effect on your life of knowing your result?

5. *Family communication, e.g.*

Do your family talk much about HNPCC? Who talks to who?

Do your family talk much about genetic testing?

Who talks about it the most in your family?

Who in your family knows the most about HNPCC?

Who in your family have you talked to about having a genetic test?

Do you think your family is a close family?

Parents:

Have you talked to your children about genetic testing?

How did you introduce the subject with your children? How do they feel about it?

After test results:

Who have you talked to in your family about your result? What was their reaction?

Is there anyone you have not talked to about it? Can you say why?

6. *Social context, e.g.*

Do you talk to other people (friends, work colleagues) about HNPCC / testing?

Are you happy to talk to them about it?

What are their views about genetic testing?

Is it helpful to talk about it / not talk about it with people outside your family?

Is it something people outside your family find interesting?

7. *Reframing, e.g.*

Have your thoughts and feelings about your

situation changed in any way since you first found out about HNPCC?
Have your thoughts and feelings about your situation changed in any way since you had your test result?

8. *Family relationships:*

Do you think that relationships within your family have changed in any way because of genetic testing?