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ORIGINAL ARTICLE

Cancer risk communication, predictive testing and management in France, Germany, the Netherlands and the UK: general practitioners' and breast surgeons' current practice and preferred practice responsibilities

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Abstract Genetic testing has its greatest public health value when it identifies individuals who will benefit from specific interventions based upon their risk. This paradigm is the basis for the use of predictive tests, such as BRCA1/BRCA2 testing which has become part of clinical practice for more than a decade. Currently predictive BRCA1/BRCA2 testing is offered to women using low, moderate and high risk based upon family history as cut-off levels. Non-genetic health professionals such as general practitioners (GPs) and breast surgeons (BS) are seen as gatekeepers to manage demand and/or facilitate access to appropriate services for high-risk patients. Data about current practices are lacking. The paper presents data on the current practice of GPs' and BS' cancer risk assessment, referral practices and preferred practice responsibilities for women at risk for familial breast cancer in France, Germany, the Netherlands and the UK derived by a self-administered questionnaire send to a representative sample of GPs and BS in the four countries. One thousand one hundred ninety-seven GPs and 1,223 BS completed the questionnaire. Both GPs and BS reported that they are consulted by a considerable number of patients presenting with concerns about a family history of cancer. Both commonalities and striking differences could be observed between GPs and BS from the four participating countries. GPs from France and Germany reported significantly higher proportions taking a family history of cancer including the extended family than GPs from the Netherlands and the UK. Most GPs from France, Germany and the Netherlands stated their willingness for providing risk assessment for an unaffected (high-risk) woman with a family history of breast cancer and the vast majority of BS from all four countries reported that they themselves would provide risk assessment for an unaffected (high-risk) woman with a family history of breast cancer. However, a substantial number of both GPs and BS would not have taken an appropriate family history for their patient failing to take into account the paternal side of the family. GPs from Germany reported a significantly lower readiness to refer a patient with a family history of a BRCA1 mutation for specialist genetic counselling when compared to the GPs from the other countries. GPs and BS from France, Germany and the Netherlands significantly less often assigned practice responsibilities to a genetic specialist as compared to the participating GPs and BS from the UK. The outcome of the study confirms the need for capability building in genetics for non-

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genetic health professionals. Using genetic risk assessment tools without a full understanding could result in missed opportunities for cancer prevention and harm patients. In order to provide best possible services for high-risk patients presenting with cancer concerns, close collaboration with clinical geneticists should become routine part of mainstream medical practice.

Introduction

Genetic testing has its greatest public health value when it identifies individuals who will benefit from specific interventions based upon their risks. This paradigm is the basis for the use of predictive tests, such as *BRCA1/2* testing which has become part of clinical practice for more than a decade. Currently in western European countries, predictive *BRCA1/2* testing is offered to women using low, moderate and high risk based upon family history as cut-off levels. There has been a substantial increase in referrals of patients with a family history of breast cancer over the past decade as professional and public knowledge of predictive genetic testing has increased and surveillance protocols and guidelines have been established (Gadzicki et al. 2011). Today non-genetic health professionals, such as general practitioners (GPs) and breast surgeons (BS), are seen as gate keepers to manage demand and/or facilitate access for high-risk women to appropriate services. However, data about their current practices are lacking.

Objectives and methods

The data presented here were derived from the international cancer risk communication study: "Cancer risk communication, predictive testing and management in the United Kingdom, France, Germany and the Netherlands (InCRisC)". InCrisC addressed the lack of data on non-genetic professionals' current management practices of patients with or at risk for familial cancer. The overall aim was to facilitate—from a European perspective—inter- and intra-country comparison with the help of a systematic survey. Taking into account that during the last decade significant efforts have been made, mainly by European scientific societies, such as the European Society for Human Genetics (ESHG)[1], and by multicentre European networking projects, such as GenEd (Genetics education: Improving non-genetic health professionals' understanding of genetic testing), and Eurogentest[2] (Genetic Testing in Europe—Network for the further development, harmonization, validation and standardization of services), to develop shared quality standards for genetic risk communication within Europe (Skirton et al. 2013; EuroGentest 2009). The InCRisC study's major objective was to obtain, for the first time, comparative European data to assess current practices of GPs' and BS' familial cancer risk communication, management of familial breast cancer and preferred practice responsibilities. The intention is to contribute to an informed international discussion on the need for non-genetic health professionals to develop capacity and capability in genetics within their clinical area in order to strengthen services' responsiveness for the health needs of those with or at risk for familial cancer.

Four countries participated: France (F), Germany (D), the Netherlands (NL) and the UK. Data were obtained by using a standardized, self-administered 70-item questionnaire mailed to a random sample of GPs (n =3,999) and BS (n =3,293)[3] in the four countries. Data on the study population, sampling methods, the construction of the questionnaire, piloting and validation protocols and response rates have already been published in detail (Den Heijer et al. 2013).

In all four countries, access to genetic testing relies basically on the family history of breast and ovarian cancer. Presented here are self-reported risk assessment practices and preferred practice responsibilities when a patient presents with cancer concerns to a GP or a BS. To the best of our knowledge, this is the first time such data are reported internationally.

Risk assessment practices and preferred practice responsibilities were measured by the following:

1. Clinical scenarios (presented as vignettes) representing Louise, an unaffected 35-year-old women whose 32-year-old sister was recently diagnosed with breast cancer and who wants to know her own risk for developing breast cancer;
2. Closed answer questions such as the following:
   - Asking the participating GPs whether they would:(1) take a cancer family history, (2) specifically ask about cancer history for first degree relatives, (3) specifically ask about cancer history for the extended family, when a patient with no personal history of cancer presents with cancer concerns (given answer categories: "never", "very rarely", "rarely", "occasionally", "frequently", "always");
   - Asking both participating GPs and BS whether they would provide risk assessment based upon the family history when an unaffected woman with a family history of breast cancer consults them about her risk

[1] See the ESHG's Public and Professional Policy Committee recommendations: https://www.eshg.org/ppc0.html (last accessed July 22, 2013)
[2] http://www.eurogentest.org (last accessed July 22, 2013)
[3] Because in France, breast surgeons who treat breast cancer can belong to different specialities such as surgery or obstetrics and gynaecology, a random sample was drawn of (a) surgeons with a practice of breast surgery and (b) obstetricians and gynaecologists. In the following, the two groups will be characterized as FGyn and FChir.

of developing breast cancer (given answer categories: "never", "very rarely", "rarely", "occasionally", "frequently", "always");

- Asking both participating GPs and BS who they think should perform the following tasks for patients with or at risk for familial breast cancer: (1) explain inheritance pattern of familial breast cancer, (2) inform about breast cancer risk for the relatives, (3) inform about breast cancer testing, (4) disclose breast cancer genetic test results, (5) provide support after breast cancer genetic testing, (6) inform about possible management options available after the results of breast cancer testing.

The vignettes and the questions were developed by the international multidisciplinary study group presenting this paper. During the pilot phase of the questionnaire, the vignettes and the questions were presented to GPs and BS in the participating countries for comments and amendments.

Provided here is the short descriptive overview on study results presented at the international symposium "Predictive Genetic Testing, Risk Communication and Risk Perception", Berlin, November 2011. The presentation focussed on commonalities and differences in regard to risk assessment practices and preferred practice responsibilities in the four countries. Bivariate analyses (chi square) for categorical data and *t* test for continuous data were provided to describe country differences using SPSS 18.0 statistical package. The level of significance used for testing was 0.05.

Results

Socio-demographic variables of the respondents

One thousand one hundred ninety-seven GPs (D: $n=450$; F: $n=275$: NL: $n=264$; UK: $n=208$) and 1,223 BS (D: $n=466$; F: $n=477$, including FGyn $n=333$ and FChir $n=144$; NL: $n=123$; UK: $n=157$) completed the questionnaire. Socio-demographic variables of the respondents varied significantly between countries and between the health professions. GPs from Germany had the highest mean age whereas GPs from the UK had the lowest mean age (mean age of GPs: D, 54.1 years; F, 48.5 years; NL, 49.4 years; UK, 45.1 years, *p* values: F/D, ≤.001; F/NL, .184; F/UK, ≤.001; D/NL, ≤.001; D/UK, .001; NL/UK, .001). BS from France had the highest mean age and BS from the Netherlands had the lowest (mean age of BS: D, 50.7 years; F, FGyn 51.3 years and Fchi, 53.9 years; NL, 46.7 years; UK, 48.3 years; *p* values: FGyn/D, .362; FChir/D, ≤.001; FGyn/NL, ≤.001; FChir/NL, .001; FGyn/UK, .001; FChir/UK, .001; NL/D, .001; NL/UK, .147; D/UK, .002). GPs from the Netherlands had the lowest percentage of females whereas the highest percentage of female GPs came from the UK (% of females GPs: D, 34.6; F, 31.7; NL, 33.8; UK, 58.7; *p* values: F/D, ≤.497; F/NL, .431; F/UK, ≤.001; D/NL, .837; D/UK, .001; NL/UK, ≤.001). FChir from France had the lowest percentage of females whereas the highest percentage was represented by the responding FGyn (% of females BS: D, 45.4; FGyn, 60.4; FChir, 10; NL, 30.8; UK, 34.0; *p* values: FGyn/D, .001; FChir/D, .001; FGyn/NL, .001; FChir/NL, .001; FGyn/UK, 001; FChir/UK, .001; NL/D, .007; NL/UK, .626).

Current practice

How often is a cancer family history raised in a consultation?

The majority of GPs from all four countries reported that a cancer family history is raised in a consultation "at least once a week"/"once a month" (when the two answer categories are taken together). The highest consultation rates were reported by the participating Dutch GPs (35.4 %, at least once a week; 48.3 %, at least once a month; taken together, 83.7 %; *p* values: NL/D, ≤.0001; NL/F,≤.0001; NL/UK, ≤.0001), the lowest rates were reported by the French GPs (22.4 %, at least once a week; 35.3 %, at least once a month; taken together, 57.7 %; *p* values: F/D, ≤.0001; F/UK, .034). The consultation rates reported by both GPs from Germany and the UK were nearly the same (D: 26.8 %, at least once a week; 40.4 %, at least once a month; taken together, 67.2 %; UK: 21.3 %, at least once a week; 44.9 %, at least once month; taken together, 66.2 %; *p* value: D/UK, .662).

Significantly higher consultation rates were reported by both the BS from the Netherlands and the UK (NL: 57.7 %, at least once a week; 35.8 %, at least once month; taken together: 93.5 %; UK: 76.9 %, at least once a week; 19.9 %, at least once month; taken together, 96.8 %, *p* value: NL/UK, .422) when compared to the consultation rates reported by the BS both from Germany and France (*p* values: NL/D, ≤.0001; NL/F, ≤.0001; UK/D, ≤.0001; UK/F, ≤.0001). In Germany, 40.2 % of the BS reported that a cancer family history is raised in a consultation at least once a week and 38.4 % reported at least once a month (taken together, 78.6 %). The BS from France reported the lowest consultation rates for both FChir (24.5 %, at least once a week; 27.3 %, at least once a month; taken together, 51.8 %) and for FGyn (13.6 %, at least once a week; 26.9 %, at least once a month; taken together, 40.5 %) compared to the rates reported by BS from Germany, the Netherlands and the UK (*p* value F/D ≤.0001).

Taking a family history: GPs

Both GPs from Germany and France, when compared to GPs from the Netherlands and the UK, reported significantly higher frequencies in taking a family history when a patient with no personal history of cancer presents with cancer

concerns. Of the GPs, 76.6 % from Germany and 74.3 % from France reported that they would always take a family history (p value D/F .449) whereas only 36.0 % of the Dutch and 40.1 % of the British GPs reported that they always take a family history (p values: D/NL, ≤.0001; F/NL, ≤.0001; D/UK, ≤.0001; F/UK, ≤.0001). Correspondingly significant differences were found between the proportion of GPs from both Germany and France when compared to the proportion of GPs from the Netherlands and the UK reporting (a) how often they specifically ask about cancer history in first degree relatives ("always": D, 78.6 %; F, 71.3 %; NL, 44.7 %; UK, 38.0 %; p values: D/F, .115; D/NL, ≤.0001; D/UK, ≤.0001; F/NL, ≤.0001; F/UK, ≤.0001) and (b) how often they specifically ask about cancer history of the extended family. ("always": D, 47.8 %; F, 41.4 %; NL, 15.5 %; UK, 13.6 %; p values: D/F, .190; D/NL, ≤.0001; D/UK, ≤.0001; F/NL, ≤.0001; F/UK, ≤.0001; NL/UK, .091).

Providing risk assessment for an unaffected woman with a family history of breast cancer: GPs and BS

The majority of GPs from all countries, reported that they provide "always"/"frequently" risk assessment (GPs—D: always, 28 %; frequently, 33.5 %; taken together, 61.5 %; F: always, 28.6 %; frequently, 39.3 %; taken together, 67.9 %; NL: always, 18.4 %; frequently, 35.5 %; taken together, 53.9 %; UK: always, 22. 7 %; frequently, 32.0 %; taken together, 51.7 %). Observed differences are significant between the GPs from France and the Netherlands (p value, .007), from France and the UK (p value, .001) and from the UK and Germany (p value, .004). Differences are not significant between the GPs from Germany and France (p value, .069), between the GPs from Germany and the Netherlands (p value, .073) and between the GPs from the Netherlands and from the UK (p value, .577).

GPs from both France and Germany tend to differ less in reported proportions of family history taking practices and reported more often taking family histories and to more frequently providing risk assessment based upon a family history than their colleagues from the Netherlands and the UK, whereas GPs from both the Netherlands and the UK tend to less often take a family history and provide risk assessment less often and reported similar proportions of risk assessment practices.

The vast majority of BS from all four countries reported that they would provide risk assessment for an unaffected woman with a family history of breast cancer. The highest proportion was reported by BS from the UK, the lowest by the BS from France ("always"/"frequently": UK, 85.3 %; NL, 80 %; D, 78 %; FGyn, 76.8 %; FChir, 76.5 %). However, the differences were not significant between the countries.

Clinical scenario

1. Louise, an unaffected 35-year-old woman whose 32-year-old sister was recently diagnosed with breast cancer wants to know her own risk for developing breast cancer:

About which other family members would you take a cancer history?

Both GPs and BS from all four countries tend to focus on female relatives when taking a family history of breast cancer and fewer are reporting that they are routinely ("always") collecting information on male relatives and on relatives on the father's side of the family (Table 1). While more than 90 % of the GPs from France, Germany, the Netherlands and the UK reported that they would collect data on a cancer history of the mother (almost 100 % in all countries), other sisters and maternal grandmother (well above 90 % in all countries) and no significant differences could be observed between the GPs from all countries, they were less likely to report collecting this information for the father, the brothers, the maternal grandfather, paternal grandmother and grandfather. This tendency could be observed in all four countries. However significant differences were found between the four countries in regard to the proportion of GPs and BS who reported that they would take the father's side of the family into account when taking a family history.

Significantly less GPs from France and Germany reported that they would take a cancer history from the father (F, 35.6 %; D, 40.9 %) and the brothers (F, 26.2 %; D, 31.3 %) as compared to GPs from the Netherlands (fathers, 64.8 %; brothers, 47 %) and the UK (fathers, 52.4 %; brothers, 39.4 %) (p values for differences for both fathers and brothers: F/NL, ≤.0001; F/UK, ≤.0001; D/NL, ≤.0001; for fathers: D/UK, .005; for brothers: D/UK, .037). The proportion of GPs from the Netherlands who reported taking a cancer history of the father is significantly higher when compared to the reported proportions by the GPs from all other countries including the UK (p value NL/UK, .008).

Comparing the responses from the BS, a similar trend could be observed (Table 1). Almost all BS from France, Germany, the Netherlands and the UK reported collecting information on a cancer history of the mother, other sisters and the maternal grandmother (no significant differences). Fewer BS reported that they would collect information on the father's side of the family. However, significant differences were found; 91.4 % of the BS from the UK and 90.2 % of the BS from the Netherlands (p value: UK/NL, .714) reported taking a cancer history of the father as compared to only 70.6 % of the BS from Germany (p value D/UK, ≤.0001; D/NL, ≤.0001) and to only 63.3 %(FGyn)/51,7 %(FChir) of the BS from France (p values: F/D, .005; F/NL, ≤.0001; F/NL, ≤.0001). A

Table 1 Clinical scenario: an unaffected 35-year-old woman has a 32-year-old sister who has recently been diagnosed with breast cancer. She wants to know about her own risk. About which family members would you take a cancer history?

	Family member	France[a] (%)	Germany (%)	Netherlands (%)	UK (%)
General practitioners	Mother	99.6	99.6	99.6	100
	Father	35.6	40.9	64.8	52.4
	Other sisters	98.2	96.9	99.6	97.6
	Brothers	26.2	31.3	47.0	39.4
	Maternal grandmother	93.5	94.4	90.9	92.8
	Maternal grandfather	19.3	30.0	37.5	30.3
	Paternal grandmother	56.7	65.6	63.6	57.7
	Paternal grandfather	17.5	27.7	33.3	24.5
Sample size (GPs)		(n=275)	(n=450)	(n=264)	(n=208)
Breast surgeons	Mother	99.6 (100.0)	99.8	100	98.0
	Father	63.3 (51.7)	70.6	90.2	91.4
	Other sisters	98.8 (98.6)	99.1	98.4	97.4
	Brothers	44.3 (37.8)	57.9	65.9	76.3
	Maternal grandmother	96.4 (95.1)	93.5	96.7	96.1
	Maternal grandfather	41.3 (34.3)	49.2	66.7	78.9
	Paternal grandmother	64.8 (66.4)	62.6	84.4	89.5
	Paternal grandfather	37.7 (35.0)	44.1	62.6	77.0
Sample size (BS)		(n=333; n=145)	(n=466)	(n=144)	(n=123)

[a] BS=F_{Gyn}, in brackets F_{Chir}

consistently higher proportion of the BS from the Netherlands and from the UK reported taking a cancer history of the brothers, maternal grandfather, paternal grandmother and grandfather as compared to the BS from France and Germany.

2. Louise's lifetime risk of developing breast cancer is approximately 20 % based upon the family history of her affected 32-year-old sister.

Would you provide cancer risk information for Louise yourself?

Most GPs from Germany (74.7 %), France (74.2 %) and the Netherlands (71.3 %) reported that they themselves would provide risk information for Louise. The GPs from the UK differed significantly. Only 21 % reported that they would provide risk information themselves for Louise (p values: D/UK, ≤.0001; F/UK, ≤.0001; NL/UK, ≤.0001). The same trend—significant differences between the UK and the other three countries—was observed for the BS. Most BS from Germany (81.1 %), France (FGyn, 75.5 %/FChir, 87.1 %) and the Netherlands (77.3 %) reported that they would provide risk information themselves whereas only 48.7 % of the BS from the UK reported that they would provide risk assessment themselves for Louise (p values: D/UK, ≤.0001; F/UK, ≤.0001; NL/UK, ≤.0001).

Would you refer Louise for specialist cancer risk assessment? (GPs)/ for specialist cancer genetic consultation? (BS)

The highest proportion of GPs who answered that they would "certainly not refer" or "probably not refer" Louise came from France (certainly not, 11.2 %; probably not, 40.5 %; taken together, 51.7 %), the lowest proportion who reported that they would "certainly not" (1.4 %) or "probably not" (10.6 %; taken together, 12.0 %/) refer Louise came from the UK (p value: F/UK, ≤.0001). Of the GPs from the Netherlands, 26.0 % reported that they would probably not refer and 1.9 % would certainly not refer (taken together, 27.9 %) and 5 % of the GPs from Germany reported that they would certainly not refer and 17.9 % probably not refer Louise (taken together, 22.9 %). From both Germany (54.9 %) and the UK (53.6 %), a majority of the GPs reported that they would certainly refer Louise, whereas only 23.3 % of the GPs from France and 26.4 % of the GPs from the Netherlands reported that they would certainly refer their high risk patient Louise ("yes, certainly", "yes probably"; taken together: UK, 88.0 %; D, 77.2 %; NL, 62.6 %; F, 48.3 %; p values for differences between all countries, ≤.0001)

The BS were asked whether they would refer Louise to a genetic cancer risk consultation. From both the Netherlands (54.0 %) and the UK (54.0 %), a majority of BS answered that they would certainly refer Louise. Taken together with the answer category "yes, probably", the reported proportions for

the Netherlands and the UK respectively are 81.3 and 73.3 % (p value: NL/UK, .306). The proportions of BS from both Germany and France who reported that they would certainly refer Louise are significantly lower (D, 37.1 %; FGyn, 45.7 %/ FChir, 39.8 %) No significant differences were found between the BS from France and Germany who responded that they would either "certainly not" (D, 7.9 %; FGyn, 9.0 %; FChir, 7.9 %) or "probably not" (D, 26.5 %; FGyn, 28.1 %/FChir, 18.5 %) refer Louise to a specialist genetic cancer consultation.

3. Six months later: Louise's cousin developed breast cancer and reportedly tested positive for a mutation in the high risk cancer gene BRCA1. Louise wants to know how this affects her risk.

Would you provide information about cancer risks based upon the likelihood of carrying the mutation?

Significant differences were observed between the proportions of GPs from the four countries who reported that they themselves would provide information about Louise's cancer risks based upon the likelihood of her carrying the mutation.

The lowest proportion (5.2 %) of GPs who reported that they would certainly provide risk information themselves was found in the UK (yes, probably, 19.1 %; probably not, 44.8 %; certainly not, 30.9 %), followed by the following: 21.7 % (yes, certainly) of GPs from Germany (yes, probably, 31.4 %; probably not, 29.6 %; certainly not, 17.2 %), 25.5 % (yes, certainly) of GPs from the Netherlands (yes, probably, 40.2 %; probably not, 23.9 %; certainly not, 10.4 %) and 49.8 % (yes, certainly) of GPs from France (yes, probably, 42.3 %; probably not, 5.3 %; certainly not, 2.6 %) (p values: F/UK, ≤.0001; D/UK, ≤.0001; NL/UK, ≤.0001; F/NL, ≤.0001; D/NL, ≤.0001; UK/NL, ≤.0001).

Only from the UK, a majority (75.7 %) of GPs reported that they would certainly not/probably not provide risk information themselves. The proportions reported by GPs from Germany (46.8 %), from the Netherlands (34.3 %) and from France (18.8 %) are significantly lower.

A higher proportion of BS from all four countries—as compared to their GP counterparts—stated that they themselves would provide cancer risk information for Louise. The highest proportion was reported by the BS from France (FGyn: yes, certainly, 63.3 %; yes, probably, 29.4 %; taken together, 92.7 %/(FChir: yes, certainly, 65.9 %; yes, probably, 29.6 %; taken together, 95.5 %) followed by the BS from the Netherlands (yes, certainly, 50.4 %; yes, probably, 29.4 %, taken together, 79.8 %) and the BS from the UK (yes, certainly, 28.7 %; yes, probably, 36.4 %; taken together, 65.1 %). The lowest proportion was reported by the BS from Germany (yes, certainly, 24.3 %; yes, probably, 37.5 %; taken together, 61.7 %). Differences between BS from Germany and the UK are not significant (p value .650). Significant differences between reported proportions were found between the BS from France/Netherlands (p value, .001), between the BS from France/Germany (p value ≤.0001), between the BS from France/UK (p value ≤.0001), between BS from the Netherlands/Germany (p value ≤.0001) and from the Netherlands/UK (p value, .001).

Would you refer for specialist genetic counselling?

Although a majority of GPs in all four countries reported that they would refer Louise for genetic counselling, significant differences exist between relative proportions. The lowest proportion of GPs who reported that they would certainly refer Louise was observed in Germany. Only 51.2 % reported that they would certainly refer Louise (other answer categories: yes, probably, 26.6 %; probably not, 16.8 %; certainly not, 6.5 %) as compared to 71.1 % from the UK (other answer categories: yes, probably, 24.0 %; probably not, 4.4 %; certainly not, 0.5 %), 68.7 % from the Netherlands (other answer categories: yes, probably, 28.6 %; probably not, 2.7 %; certainly not, 0.0 %) and 65.1 % from France (other answer categories: yes, probably, 24.0 %; probably not, 7.4 %; certainly not, 1.8 %). The differences between the GPs from Germany and the GPs from the other countries are significant (p values: D/FR, ≤.0001; D/NL, ≤.0001; D/UK, ≤.0001).

The majority of BS from all four countries reported that they would certainly refer Louise for genetic counselling. These proportions are higher than the proportions of GPs who would certainly refer in the respective countries. However—albeit from a high starting level—the proportion of BS from Germany reporting certain referral ("yes, certainly" 71.9 %, responses other answer categories: yes, probably, 20.8 %; probably not, 5.4 %; certainly not, 2.0 %) is significantly lower than the proportion of BS from France (FGyn: "yes, certainly" 83.0 %, responses other answer categories: yes probably, 14.3 %; probably not, 2.1 %; certainly not, 0.6 %/FChir: yes, certainly, 87.5 %; yes, probably, 13.6 %; probably not, 0.0 %; certainly not, 0.0 %), the proportion from the Netherlands (yes, certainly, 86.9 %; yes, probably, 12.3 %; probably not, 0.8 %; certainly not, 0.0 %), and the proportion from the UK (yes, certainly, 89.5 %; responses other answer categories: yes, probably, 7.8 %; probably not, 2.6 %; certainly not, 0.0/p values: D/F, .003; D/NL, .004; D/UK, ≤.0001). There are no significant differences between the proportions reported for referrals by the BS from France, the Netherlands and the UK.

Preferred practice responsibilities

Preferred practice responsibilities: GPs

Table 2 provides an overview on the responses for six preferred practice responsibilities reported by the participating

Table 2 GPs: For patients with or at risk for familial breast cancer. Who do you think should undertake the following tasks?

%

Should be done by…		Myself	A genetic specialist	A breast specialist
(a) Explain the inheritance pattern of familial breast cancer	France	63.6	26.5	9.6
	Germany	30.0	44.8	25.2
	Netherlands	49.7	43.5	6.8
	UK	33.8	60.4	5.8
p values: D-F, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F-NL, ≤.0001; F-UK, ≤.0001; NL-UK, ≤.0001				
(b) Inform about breast cancer genetic risks for relatives	France	77.7	14.3	8.0
	Germany	43.9	33.0	23.1
	Netherlands	34.5	52.1	6.1
	UK	25.8	69.8	4.4
p values: D-F, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F-NL, ≤.0001; F-UK, ≤.0001; NL-UK, ≤.0001				
(c) Inform about breast cancer genetic testing	France	56.2	29.6	14.2
	Germany	46.6	30.4	23.0
	Netherlands	41.7	54.4	3.9
	UK	41.6	53.6	4.8
p values: D-F, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F-NL, ≤.0001; F-UK, ≤.0001; NL-UK, ≤.583				
(d) Disclose the breast cancer genetic test results to the patient	France	23.5	59.3	17.2
	Germany	43.7	32.4	23.9
	Netherlands	11.6	87.3	1.1
	UK	16.9	76.7	6.4
p values: D-F, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F-NL, ≤.0001; F-UK, ≤.0001; NL-UK, ≤.003				
(e) Provide support after breast cancer genetic testing	France	86.1	5.8	8.1
	Germany	59.6	14.3	26.1
	Netherlands	66.4	28.2	5.4
	UK	57.2	34.5	8.3
p values: D-F, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F-NL, ≤.0001; F-UK, ≤.0001; NL-UK, ≤.002				
(f) Inform about possible management options available after the results of breast cancer genetic testing	France	51.5	19.0	29.5
	Germany	41.4	20.5	38.1
	Netherlands	33.4	37.9	28.7
	UK	18.4	30.9	50.7
p values: D-F, ≤.083; D-NL, ≤.0001; D-UK, ≤.0001; F-NL, ≤.0001; F-UK, ≤.0001; NL-UK, ≤.0001				

Sample size: France, $n=275$; Germany, $n=450$; Netherlands, $n=264$; UK, $n=208$

GPs. Although the GPs from the four countries differ significantly in their preferred practice responsibilities, distinct country patterns can be observed. Agreement can be found among the majority of GPs from all four countries that their practice responsibility should *be* "to provide support after breast cancer testing". However, the proportions vary significantly. The highest proportion, 86.1 %, is reported by the GPs from France, the lowest, 57.2 %, by the GPs from the UK. Unanimously, the majority of the GPs from three countries: Germany, the Netherlands and the UK considered this to be the only task out of the six that falls under their responsibility. The majority of the responding GPs from France differed from the GPs from all other countries. They ascribed most practice responsibilities (five out of six tasks) to themselves including the following tasks: "explain the inheritance pattern of familial breast cancer" (63.6 %); "inform about breast cancer genetic risk for the relatives" (77.7 %); "inform about breast cancer genetic testing" (56.2 %), "provide support after breast cancer genetic testing" (86.1 %) and "inform about possible management options available after the results of breast cancer genetic testing" (51.5 %). Only one task was considered by the majority of GPs from France to be in the realm of a genetic specialist: "disclose breast cancer genetic test results to the patient" (59.3 %). In stark contrast, the majority of GPs from the UK assigned practice responsibilities for most of the tasks (four out of six tasks) to a genetic specialist including: "explain the inheritance pattern of familial breast cancer" (60.4 %), "inform about breast cancer risk for the relatives" (69.8 %), "inform about breast cancer testing" (53.6.%) and "disclose breast cancer genetic test results to the patient"

(76.7 %). In addition, the majority (50.7 %) of the GPs from the UK reported that one task should be undertaken by a breast specialist ("inform about possible management options available after the results of breast cancer genetic testing"). The responding GPs from Germany differ from the GPs from other countries as none of the six tasks was clearly (=by the majority) assigned by them to a genetic specialist. More often than the GPs from the other countries about one in four of the GPs from Germany considered breast specialists' practice responsibilities for the six tasks.

Overall, taking the responses from the GPs from all countries into account there is only one single task: "disclose breast cancer genetic test results to the patient" on which the majority of GPs (from three countries France, the Netherlands and the UK) unanimously agreed that it should be undertaken by a genetic specialist. There are two out of six tasks including "provide support after breast cancer genetic testing" and "inform about possible management options available after the results of breast cancer genetic testing", on which the majority of the GPs from all four countries agreed that these tasks are not the practice responsibility of a genetic specialist. Otherwise significant differences could be observed between GPs from the four countries in ascribing practice responsibilities.

Preferred practice responsibilities: BS

The majority of the BS from all countries agreed on the practice responsibility for two tasks (Table 3). They agreed that "inform about possible management options available after the results of breast cancer genetic testing" should be their practice responsibility (D, 79.2 %; FGyn, 50.3 %/FChir, 63.4 %; NL, 74.9 %; UK, 62.1 %) and they agreed—just like the GPs—that the task "disclose breast cancer genetic test results to the patient" should be undertaken by a genetic specialist (D, 63.5 %; FGyn, 88.2 %/FChir, 79.3 %; NL, 94.1 %; UK, 94.1 %), although significant differences exist in regard to reported proportions. As seen above ("Preferred practice responsibilities: GPs"), BS respondents from the UK more often assigned practice responsibilities (five out of six tasks) to a genetic specialist as compared to the respondents from Germany, France and the Netherlands. The five tasks assigned by the BS from the UK to a genetic specialist include the following: "explain the inheritance pattern of familial breast cancer" (66.7 %), "inform about breast cancer genetic risk for the relatives" (74.8 %), "inform about breast cancer genetic testing" (77.8 %), "disclose breast cancer genetic test results to the patient" (94.1 %) and "provide support after breast cancer genetic testing" (61.8 %). Comparing the responses for practice responsibilities of the GPs and the BS from the UK, there was an apparent agreement between them on practice responsibilities to be assigned to a genetic specialist. The only exception was that the majority of the BS ascribed the task "to provide support after breast cancer testing" to a genetic specialist as well.

In contrast the majority of the BS subgroup (FGyn) from France assigned only one practice responsibility to a genetic specialist ("disclose breast cancer genetic test results to the patient") and claimed four practice responsibilities for their speciality including the following: "inform about breast cancer genetic risk for the relatives" (FGyn, 54.1 %/FChir, 75.3 %), "inform about breast cancer genetic testing" (FGyn, 56.9 %/FChir, 43.5 %), "provide support after breast cancer genetic testing" (FGyn, 73.0 %/FChir, 46.4 %) and "inform about possible management options available after the results of breast cancer genetic testing" (FGyn, 50.3 %/FChir, 63.4 %).

Otherwise, significant differences between the reported practice responsibilities in the participating countries were found. BS from the Netherlands were the only ones of whom a majority (66.4 %) ascribed to their specialty the task "explain the inheritance pattern of familial breast cancer" A majority of BS from France (FGyn, 54.2 %/FChir, 65.2 %) and the Netherlands (56.8 %) ascribed the task "inform about breast cancer genetic risk for the relatives" to their speciality: The BS from the UK differed significantly in the assignment of practice responsibilities (five out of six tasks) to a genetic specialist as compared to the BS from France (FGyn, one out six tasks/FChir, two out of six tasks), Germany (three out of six tasks) and the Netherlands (two out of six tasks).

Participating GPs and BS from the UK seemed to be more inclined to assign practice responsibilities to a genetic specialist than the GPs and BS from France, Germany and the Netherlands who more often preferred to assign practice responsibilities to their own specialty.

In conclusion

Applications of predictive genetic testing in clinical practice are steadily increasing for more than a decade and potential applications are expected to increase exponential due to increased speed and decreasing costs of genomic sequencing (Schmidtke et al. 2005; Department of Health 2012; Burton et al. 2012). The challenge that arises from this development is that increasingly a wider set of individuals affected by or at risk of a genetic disorder or distinct genetic pathologies are presenting to GPs and to a wide range of clinical specialities. Both the generalists and specialists need to be skilled in recognizing inherited genetic disorders, in identifying those at increased risk and in managing patient and families affected by these conditions.

Results of the InCrisC study confirm observational evidence that in today's practice GPs and BS are consulted by a considerable number of patients presenting with concerns about a family history of cancer.

Table 3 BS: For patients with or at risk for familial breast cancer: Who do you think should undertake the following tasks?

%

Should be done by…		Myself	A genetic specialist	Other specialist
(a) Explain the inheritance pattern of familial breast cancer	France[a]	49.4 (47.1)	49.7 (46.4)	0.9 (6.5)
	Germany	26.8	71.7	1.5
	Netherlands	66.4	32.8	0.8
	UK	30.7	66.7	2.6
p values: D-F_{Chir}, ≤.00011; D-F_{Gyn}, .00011; D-NL, ≤.0001; D-UK, ≤.004; F_{Chir}-NL, ≤.0001; F_{Gyn}-NL, ≤.003; F_{Chir}-UK, ≤.0001; F_{Gyn}-UK, ≤.0001; NL-UK, ≤.004				
(b) Inform about breast cancer genetic risks for relatives	France[a]	54.2 (65.2)	44.6 (26.8)	1.2 (8.0)
	Germany	43.8	55.1	1.1
	Netherlands	56.8	43.2	0.0
	UK	23.2	74.8	2.0
p values: D-F_{Chir}, ≤.0001; D-F_{Gyn}, ≤.010; D-NL, ≤.0001; D-UK, ≤.0001; F_{Chir}-NL, ≤.0001; F_{Gyn}-NL, ≤.004; F_{Chir}-UK, ≤.0001; F_{Gyn}-UK, ≤.0001; NL-UK, ≤.0001				
(c) Inform about breast cancer genetic testing	France[a]	56.8 (43.5)	42.6 (50.7)	0.6 (5.8)
	Germany	66.3	32.2	1.5
	Netherlands	40.3	59,7	0.0
	UK	20.2	77.8	2.0
p values: D-F_{Chir}, ≤.0001; D-F_{Gyn}, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F_{Chir}-NL, ≤.0001; F_{Gyn}-NL, ≤.010; F_{Chir}-UK, ≤.0001; F_{Gyn}-UK, ≤.0001; NL-UK, ≤.0001				
(d) Disclose the breast cancer genetic test results to the patient	France[a]	9.7 (15.8)	88.2 (79.2)	2.1 (5.0)
	Germany	34.9	63.6	1.5
	Netherlands	4.2	94.1	1.7
	UK	3.3	94.1	2.6
p values: D-F_{Chir}, ≤.0001; D-F_{Gyn}, ≤.0001; D-NL, ≤.0001; D-UK, ≤.0001; F_{Chir}-NL, ≤.013; F_{Gyn}-NL, ≤.359; F_{Chir}-UK, ≤.002; F_{Gyn}-UK, ≤.088; NL-UK, ≤.460				
(e) Provide support after breast cancer genetic testing	France[a]	73.0 (46.4)	17.3 (24.3)	9.7 (29.3)
	Germany	79.3	16.1	4.6
	Netherlands	48.7	47.1	4.2
	UK	29.6	61.8	8.6
p values: D-F_{Chir}, ≤.0001; D-F_{Gyn}, ≤.004; D-NL, ≤.0001; D-UK, ≤.0001; F_{Chir}-NL, ≤.0001; F_{Gyn}-NL, ≤.0001; F_{Chir}-UK, ≤.0001; F_{Gyn}-UK, ≤.0001; NL-UK, ≤.0001				
(f) Inform about possible manage- ment options available after the results of breast cancer genetic testing	France[a]	63.5 (50.3)	33.2 (36.2)	3.3 (13.5)
	Germany	79.3	17.2	3,5
	Netherlands	74.8	23.5	1.7
	UK	62.1	35.3	2.6
p values: D-F_{Chir}, ≤.0001; D-F_{Gyn}, ≤.0001; D-NL, ≤000; D-UK, ≤.0001; F_{Chir}-NL, ≤.0001; F_{Gyn}-NL, ≤.065; F_{Chir}-UK, ≤.0001; F_{Gyn}-UK, ≤.0001; NL-UK, ≤.0001				

Sample size: France subgroups: F_{Gyn}, $n=333$, F_{Chir}, $n=145$; Germany, $n=466$; Netherlands, $n=144$; UK, $n=123$

[a] France subgroup F_{Gyn} presented in brackets

Both commonalities and striking differences in the management of such patients and in preferred practice responsibilities could be observed between the GPs and BS from the four countries participating in the survey. GPs from France and Germany reported significantly higher proportions taking a family history, including first degree relatives and the extended family than GPs from both the Netherlands and the UK. No differences were found between participating BS from all four countries who reported that they would routinely provide risk assessment for an unaffected woman with a family history of cancer. However, as the data confirm, it is not a given that participating BS and GPs will take an appropriate three-generation family history for patients for whom they are willing to provide risk assessment. Data show that a substantial amount of participating GPs and BS failed to take into account paternal cancer history or cancer history of the

maternal grandfather. With the exception of participating GPs from the UK, most participating GPs from France, Germany and the Netherlands stated a readiness for providing risk assessment themselves for an unaffected high-risk (20 % risk) patient for whom a substantial number would not have taken an appropriate three-generation family history. Failing to realize paternal inheritance pattern severely impacts the provision of accurate risk assessment and could result in missed opportunities for cancer prevention and in compromised informed decision making. The data of the InCrisC study confirm findings from a Canadian study (McCuaig et al. 2010; McCuaig et al. 2011) that non-genetics health professionals tend to under evaluate the paternal or father's side of familial cancer. The findings of the InCrisC survey indicate that this may be a problem ubiquitous in many countries, irrespective of different health care systems. The data highlight the current need for capacity and capability building in genetics focused on building skills and knowledge across the health care workforce in the participating countries.

The "new genetics" is a rapidly evolving science and a relatively new field in medicine. Its associated specialty clinical genetics is a relatively young and small specialty that provides services for individuals and families affected by or at risk for an inherited disorder. Services and competences in clinical genetics include risk assessment, diagnostics, genetic testing, genetic counselling and support provided for patients and the extended family. Previous reports from the Genedconsortium (Challen et al. 2005; Schmidtke et al. 2006; Julian-Reynier et al. 2008; Benjamin et al. 2009; Nippert et al. 2011) indicate that in European countries the inclusion of genetics within undergraduate and postgraduate curricular across different non-genetics' health professionals is inconsistent and lacking in coherence.

It may be that professionals who have been trained and accredited for some time are not necessarily aware of clinical genetics' core competences or that such core competences appear to be somewhat opaque. This may explain why many participating GPs especially from France, Germany and from the Netherlands were less ready to refer a high risk patient for specialist cancer risk assessment and preferred providing risk assessment themselves. It may also explain why preferred practice responsibilities were—with the exception of GPs and BS from the UK—assigned by a majority of GPs and BS to themselves rather than to a genetic specialist. GPs and BS from the UK stand out in their willingness to assign practice responsibilities to a genetic specialist as compared to GPs and BS from the other participating countries. This may partly be explained by the GPs' explicit gatekeeper role within the National Health Service (NHS) and a more clear-cut division of tasks among clinical specialties within the NHS. In addition, health policy makers in the UK, including the Royal College of Physicians, have taken a positive, pro-active stance towards "the new genetics". Acknowledging the difficulties for practicing physicians to keep up with rapid advances in genetics, the Royal College of Physicians has a tradition of offering education programmes in genetics and the NHS has established a National Genetics Education and Development Centre. Arguably such actions may have contributed to giving clinical genetics' competences a clearer visibility among the other specialties within the NHS and may have helped to establish patient- and clinical pathways for care and referrals.

In respect to the findings from Germany, for example, GPs' reported relatively low willingness to refer an unaffected high-risk patient with a family history of a *BRCA1* mutation for specialist genetic counselling, and that none of the practice responsibilities included in the survey was clearly ascribed to a genetic specialist by GPs, it has to be kept in mind that the InCrisC survey has been conducted in 2008–2009. This was before the enactment of the *Gendiagnostik-Gesetz* (Gene Diagnostics Act) in 2009 and before the independent national *Gendiagnostik-Kommission* (Genetic Diagnostics Committee) laid out in 2011—as required by the act—binding guidelines for the provision of genetic testing services including the provision of genetic counselling (Richtlinien der Gendiagnostik-Kommission am Robert Koch-Institut 2011). It remains to be seen to what extent the recently implemented guidelines will change referral patterns for genetic counselling and will alter the perception of practice responsibilities in the future.

The outcome of the InCrisC study clearly confirms the need for capacity and capability building in genetics for health professionals outside genetics, especially in relation to the rapid development of genetic testing technologies expected to become relevant for a wide range of clinical specialties. However, acknowledging current skill gaps, embedding genetic knowledge and skills in the "real world" of daily(mainstream) medical practice, represents quite a challenge in most countries and may at best be a gradual process that needs to take into account the particular competences and needs of generalists and specialists. Probably, some type of accreditation may be also needed in order to assure quality of training and acquired knowledge skills. Using genetic information tools without a full understanding, as reported by quite a substantial proportion of the InCrisC survey participants, will prove to be detrimental for patients and their families. Above all, close collaboration with clinical genetics needs to become a routine part of mainstream medical practice, allowing full use of the experience and expertise of genetics health professionals in order to provide optimum services for patients and their families.

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