Equal access to breast cancer genetic counseling and testing

Development and implementation of a health literacy training program for surgical oncologists and specialized nurses

Jeanine van der Giessen - van der Doelen

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CHAPTER 1

General introduction

Hereditary breast cancer

Breast cancer, the most commonly diagnosed type of cancer, affects about 12% of women worldwide.[1] In the European Union, breast cancer accounts for 29% of all cancers in women, with a higher observed incidence in the more affluent countries, usually the ones with a longer established Western lifestyle.[2,3]

Approximately 5-10 % of women with breast cancer have a genetic predisposition and carry a mutation in a breast cancer gene. It is important to identify these patients because they are faced with therapeutic consequences, as well as decisions on possible preventive therapies.[4] Most inherited cases of breast cancer are associated with mutations in the *BRCA1* and *BRCA2* genes. Carriers of a *BRCA1* or *BRCA2* gene mutation have a 60-80% risk of developing breast cancer and a 10-45% risk of developing ovarian cancer. *BRCA1*/2 mutation carriers diagnosed with breast cancer have a risk of developing contralateral breast cancer that can be as high as 60%.[5] This risk is higher for women diagnosed with breast cancer before the age of 50.[6-8] Relatives are also at risk of carrying the mutation and may have an increased risk of developing cancer.

Breast cancer genetic counseling and testing

For patients diagnosed with breast cancer, it is important to identify if they meet the criteria for referral to breast cancer genetic counseling and testing. A medical consultation, were eligibility for referral is assessed, is the first step for access to genetic testing. Based on international guidelines, background characteristics of patients and information about family cancer history, a healthcare professional (mostly a surgical oncologist) verifies whether referral to breast cancer genetic counseling is appropriate.[9] Because of treatment implications for the patient and the medical implications for their relatives, it is important that eligible patients are referred for genetic counseling and DNA-testing by their treating healthcare professional. Genetic counseling helps people understand and adapt to the medical, psychological and familial implications of genetic contributions to the disease.[10] Referral to genetic counseling and testing for high-risk breast cancer patients is crucial and should preferably be offered early after diagnosis to guide treatment decisions. Because of the increased risk of contralateral breast cancer, breast cancer patients with a *BRCA1*/2 gene mutation can decide whether or not to opt for bilateral mastectomy as primary surgery. Also, risk reducing salpingo-oophorectomy is advised to *BRCA* mutation carriers, and the chemotherapeutic approach for these patients can be different.[4,11] Healthcare professionals (surgical oncologists, medical oncologists and specialized nurses) should identify patients at risk of carrying a mutation in a breast cancer gene, inform them about genetic testing and refer them for genetic counseling and testing.

Communication with breast cancer patients

Communication in cancer care is challenging, because information about treatment options has to be discussed while patients are mostly overwhelmed and distressed by fear and uncertainty.[12,13] Patients attending breast cancer care furthermore receive care from various (healthcare) professionals including surgeons, radiologists, pathologists, medical oncologists, radiation oncologists, plastic/reconstructive surgeons, primary care physicians, nurses, social workers, patient advocates, and clinical geneticists, which increases the risk of miscommunication and contradicting information.[14] On top of that, patients have to make difficult decisions about the best treatment or screening option, like participating in genetic testing. In the last decade, the increased focus on patients' perspective and patient-centered care has required a shift in communication; with enhanced patient participation in decision-making becoming increasingly important.[15] Healthcare professionals were encouraged to involve their patients in decision-making and to provide information in a way their patients understood. It has been identified as an important element in good, advanced cancer care.[16] It corresponds to the World Health Organization's (WHO) new definition of health, which employs a more active patient role, emphasizing the ability to adapt and self-manage in the face of social, physical, and emotional challenges.[17] The focus of this new definition is on patients' ability to manage their own health; they are no longer seen as passive recipients of care, but are invited to participate actively in communication with their healthcare professional. To support breast cancer patients in this process, they need access to accurate and understandable information about treatment modalities and referral to cancer genetic counseling and testing (including risk estimation).[18] They need effective patient-centered communication, i.e. communication that is tailored to their level of understanding. This is a prerequisite to understand their diagnosis, prognosis, treatment and screening options. Adapting information to patients' skills, needs and preferences is also important to enhance their decision-making, recall of information and satisfaction.[19]

Disparities in access to breast cancer genetic counseling and testing

Although genetic counseling and testing is clinically relevant for all high-risk patients with breast cancer, patients with a lower level of education and migrant patients seem to have poorer access to cancer-related genetic counseling and testing.[20-24] Due to various physician- and patient-related factors, not all patients eligible for genetic counseling and testing are referred by their physicians.[25,26] Sometimes underinsurance or fear of uncovered costs play a role,[27] but more often non-financial barriers like lower education, limited language proficiency, cultural diversity or a lower level of health literacy are contributing factors.[21,23,24,28] Access barriers continue to limit the use of genetic counseling and testing for these groups of patients, indicated as communication-vulnerable patients. Therefore they do not receive the maximal efficacy of cancer prevention strategies.

Barriers and facilitators in referral to breast cancer genetic counseling and testing

We developed a conceptual framework which describes potential barriers and facilitators of referral to breast cancer genetic counseling, derived from scientific literature, including our previous work. The framework is based on the ASE model, the health belief model as well as on causal pathways of the impact of health literacy on medical communication and access to care.[29-31] The ASE model has general scientific acceptance and explains behavior by linking attitude, social norm and self-efficacy with behavior and behavioral intention.[30] The health belief model is a theoretical model that is used to explain and predict individual changes in (health) behaviors.[32] Figure 1 describes factors influencing referral to breast cancer genetic counseling. Effective communication, as well as physician recommendation are important factors influencing referral to breast cancer genetic counseling.[23,26,33] The actual uptake of genetic testing can be influenced by several psychological factors like cancer worry, perceived barriers, as well as family influence and media exposure, and there is further evidence that low uptake rates are more likely to occur in population groups with lower education, lower income or in those with a migrant background.[34-38] In medical communication about (referral) to breast cancer genetic counseling and testing, patient-related factors as well as healthcare professional-related factors play a role.

Patient-related factors

Socio-demographic background characteristics of patients, such as gender, age, socio-economic position and educational level, influence access to care in general and also appear to matter in the referral to (breast) cancer genetic counseling. Moreover, a lower level of education is linked to lower use of genetic counseling and testing services; such patients show less interest in cancer genetic counseling and testing and take less initiative for referral.[22] In breast cancer care, migrant status is also associated with a lower referral rate in genetic counseling and testing.[14,21,22,24,25,40] Also, the younger age-group of patients with a non-western background seems to be underrepresented.[39]

The lower uptake of genetic counseling and testing of patients with a lower level of education or a migrant background, may also be affected by the level of health literacy. Health literacy is broadly recognized as a critical factor affecting communication in cancer care and several studies indicate that health literacy plays an important role in explaining health disparities.[40-45,46] It is generally defined as a persons' ability to access, understand, appraise and apply health information to make a decision to maintain and improve health, although the literature on health literacy provides no unanimously accepted definition of the concept.[41] In the Netherlands, 29% of Dutch adults have a low or a limited level of health literacy.[47] Health literacy has a potential role in explaining health disparities and the World Health Organization (WHO) therefore considers health literacy to be a central determinant of health inequalities. According to the American Medical Association, limited health literacy is a stronger predictor of a person's health than individual factors such as age, income, employment status, education level or race.[48] Low education is associated with low

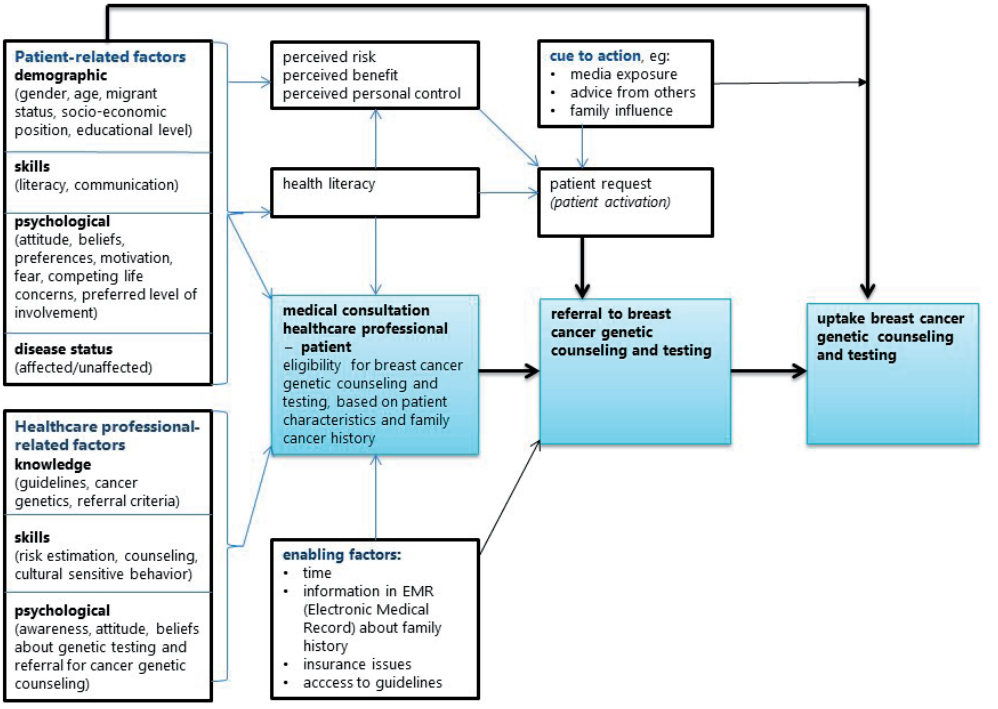

Figure 1. Framework referral to breast cancer genetic counseling and testing.

health literacy and health literacy plays a larger role among those with lower education than among those with higher education.[49] Also, migrants show increased vulnerability to a lower level of health literacy and are at risk of health disparities and significantly worse health outcomes.[49,50] Individuals with limited health literacy may understand less from written and oral communication about (genetic) information and may engage less in discussions with healthcare professionals.[51-54] This compromises their ability to make informed health decisions and has been linked to many poor health outcomes.[55] Limited health literacy, and also limited numeracy generally complicate shared decision-making and the level of health literacy has, in some studies, been suggested as a barrier to participation in cancer screening programs.[16,56-58] Patients with limited health literacy also show a decreased patient-initiated inquiry into genetic counseling and testing.[59] This lack of initiative matters, since physicians are more likely to order a genetic test if patients inquire about genetic testing.[60,61] A focus on health literacy may therefore be a useful strategy for reducing disparities in referral to breast cancer genetic counseling and testing.

Healthcare professional-related factors

While an adequate level of patients' communication skills is necessary for reducing disparities in access to breast cancer genetic counseling and testing, the way in which information is communicated by healthcare professionals also plays a role. Physicians have to estimate

breast cancer patients' risk of carrying a cancer gene mutation in conformity with the latest referral criteria. They estimate patient eligibility for genetic counseling and testing on patient characteristics (e.g. age at diagnosis, and tumor characteristics) and family cancer history.[25] Healthcare professionals in oncology, i.e. surgical oncologists and specialized nurses, are main referrers to breast cancer genetic counseling and testing; lack of a recommendation for referral is an important reason for not testing.[21,23,26,39] Their patients, receiving a referral, generally intend to undergo genetic counseling.[62] Failure to identify and refer women at risk may be ascribed to the lack of time to assess a full familial cancer history, but even more to ineffective communication.[63,64] As various studies show, referral is not always adequately discussed with patients with limited level health literacy or a migrant background.[26,65,66] For effective referral, healthcare professionals need to address patients' level of health literacy and adapt their communication accordingly, but most have a limited understanding of (the consequences of) health literacy.[67] This lack of awareness and understanding restricts their ability to communicate effectively with patients with limited health literacy. Among healthcare professionals employing effective communication techniques is important in reducing health disparities.[68] Communication skills training can improve healthcare professionals' abilities and levels of confidence in effectively communicating with patients with limited health literacy or a migrant background, and has previously been shown to produce a significant and durable increase in the self-efficacy when communicating with these groups of patients.[69-72]

As described in our framework, medical consultation, i.e. discussing referral to breast cancer genetic counseling and testing, is an important first step in the referral process. However, it can only proceed well when healthcare professionals are helped to recognize patients with limited health literacy and to learn more effective ways to communicate about genetic counseling and testing.[67] Communication skills training has previously been shown to produce a significant and durable increase in the self-efficacy of healthcare professionals to communicate effectively.[69,70,72] A health literacy training program, tailored to the context of genetic counseling and testing, might contribute to more effective communication and to an increase of referral to breast cancer genetic counseling and testing of patients with limited health literacy or a migrant background.

Aim of this thesis

The general aim of the research project described in this thesis was to develop, implement and evaluate an intervention to reduce disparities in referral to breast cancer genetic counseling and testing and to increase the uptake for communication vulnerable patients. Within the Erfo4all project, we developed a health literacy training program (Erfo4all) for healthcare professionals (oncological surgeons and specialized nurses) in order to improve their knowledge, awareness and self-efficacy in communication with patients with a limited health literacy or a migrant background and implemented it in three regions in the Netherlands.

Specific research questions were:

1. What are the differences in educational level and migrant status between counselees referred for (breast) cancer genetic counseling and the general Dutch population?
2. How can we systematically develop a health literacy training program for healthcare professionals and a plain-language guide to improve communication about breast cancer genetic counseling and testing with patients with limited health literacy or a migrant background?
3. How do healthcare professionals perceive the usefulness and acceptability of the health literacy training program and the plain-language guide?
4. What is the impact of the health literacy training program on healthcare professionals' knowledge, awareness and self-efficacy in communicating with patients with limited health literacy or a migrant background?
5. What is the impact of the training program on referral of patients with limited health literacy or a migrant background to breast cancer genetic counseling and testing?

Outline of the thesis

This thesis is divided into three parts. The first part focuses on participation rates of patients with a lower level of education or a migrant background in cancer genetic counseling and testing and the problem of underreferral. Within this part, *chapter two* describes the results of our study conducted to determine personal characteristics and demographics of those referred to cancer genetic counseling.

The second part of this thesis describes a health literacy training program and a plain-language guide to improve communication about breast cancer genetic counseling and testing. In it, *chapter 3* describes the systematic development and the evaluation of a training program for healthcare professionals (i.e. oncological surgeons and specialized nurses) to improve communication about breast cancer genetic counseling and testing with patients with limited health literacy. *Chapter 4* shows the development of a plain-language guide for breast cancer genetic counseling and testing designed to assist patients with limited health literacy to make informed decisions to participate in genetic testing.

The third part of this thesis describes the evaluation of the training program and its effect on disparities in referral to breast cancer genetic counseling and testing. In it, *chapter 5* reports on the extent to which the training program met its goals. *Chapter 6* describes the results of a multicenter study on the rates of referral to breast cancer genetic counseling and testing with, as primary outcome, the relative number of patients with limited level of health literacy or a migrant background before and after the health literacy intervention. In *chapter 7,* the main results are summarized, followed by a reflection on the methodology and implications for daily clinical practice and future research.

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PART I

Disparities in referral to (breast) cancer genetic counseling and testing

CHAPTER 2

2

Referral to cancer genetic counseling: do migrant status and patients' educational background matter?

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Abstract

Participation rates in cancer genetic counseling differ among populations, as patients with a lower educational background and migrant patients seem to have poorer access to it. We conducted a study to determine the present-day educational level and migrant status of counselees referred to cancer genetic counseling. We assessed personal characteristics and demographics of 731 newly referred counselees. Descriptive statistics were used to describe these characteristics. The results show that about 40% of the counselees had a high educational level and 89% were Dutch natives. Compared to the Dutch population, we found a significant difference in educational level ($p =< 0.01$) and migrant status ($p =< 0.001$). This suggests disparities in cancer genetic counseling and as a result of that, suboptimal care for vulnerable groups. Limited health literacy is likely to pose a particular challenge to cancer genetic counseling for counselees with a lower education or a migrant background. Our study points to considerable scope for improvement in referring vulnerable groups of patients for cancer genetic counseling.

Keywords: cancer genetic counseling; referral; migrant status; educational level.

INTRODUCTION

For families in which a hereditary form of cancer is suspected, cancer genetic counseling can add to the optimal treatment and clinical management of patients. It can serve as a valuable aid for surveillance and/or preventive surgery for patients affected by cancer and their unaffected family members. Therefore, identifying patients eligible for cancer genetic counseling and referring them is important. Unfortunately, due to various physician- and patient-related factors (Brandt et al. 2008), not all patients eligible for genetic counseling are recognized by their physicians (Kurian et al. 2017).

Physician-related factors

From physicians' perspective, patient eligibility for cancer genetic counseling is based on family history, patient cancer history and patient request (Brandt et al. 2008). The first challenge for physicians is to identify patients eligible for referral by gathering adequate information about the family history. As shown, EMRs (electronic medical records) do not always contain enough information about family history (Sollie et al. 2016; Vogel et al. 2012). Vogel et al (2012) found that only 50% of the patients who are eligible for referral for Lynch syndrome and hereditary breast- and ovarian cancer could be identified by the EMR. Current standard clinical practices seem insufficient at identifying patients who meet the criteria for referral to genetic counseling.

The majority of physicians experience a lack of time to collect detailed information about family history (Al-Habsi et al. 2008; Wood et al. 2008). Insufficient knowledge about hereditary cancer and about the criteria for referral acts as another barrier for referral (Dekker et al. 2013; Panic et al. 2014). Besides, physicians tend to overestimate the risk of patients who actually are at population risk, while underestimating risk of patients who are at increased risk of developing cancer (Baldwin et al. 2014). Despite the fact that referral guidelines are sufficiently available, numerous studies have shown that physicians are still lacking knowledge of genetics and the latest criteria for referral (Douma et al. 2016; Prochniak et al. 2012).

When physicians are convinced there's a high probability that the risk is hereditary, they have a tendency to refer patients for periodic screening examinations instead of genetic counseling (Burke et al. 2009; Sollie et al. 2016). For colorectal cancer syndromes, Prochniak et al. (2012) found that, although physicians endorsed guidelines as a significant influence on their practice decisions, these guidelines did not influence the referral to cancer genetic counseling. The low adherence to guidelines for referral and poor registration of family history may also be responsible for the differences in referral to cancer genetic counseling in migrant patients. In a study among Turkish and Moroccan patients by Baars et al. (2016), the lowest referral rates are observed in the group of women with breast cancer at young age, despite the fact that age < 40 years is a criterion for referral, independent of family history and migrant status (Baars et al. 2016).

Patient-related factors

Patient characteristics influence the referral process as well. Patients' request for cancer genetic counseling and their concerns about family members are important determinants for cancer genetic counseling. Van Riel et al. (2012) showed that the majority of counselees initiate a referral themselves (van Riel et al. 2012), while Brandt et al. (2008) found that 73% of a group of primary care physicians and specialists base patient eligibility for referral on patient request and 54% did not refer eligible patients due to patient disinterest. When physicians are not sure whether screening is recommended, patients who expect screening and those who are more anxious, are more often referred (Haggerty et al. 2005). Apparently, cancer related concerns, but also perceived cancer risk and the belief that family history influences cancer risk, contribute to referral for cancer genetic counseling (Bellcross et al. 2015).

Lack of awareness and/or knowledge about personal risk, medical history and genetic services, seem to act as barriers to referral to cancer genetic counseling (Delikurt et al. 2015). Research from Allford et al. (2014) suggests low awareness and understanding of familial cancer risk among minority ethnic communities (Allford et al. 2014). Also socio-cultural variations in beliefs, notably stigma about cancer of inherited risk of cancer, have been identified. For migrant breast cancer patients, language difficulties and lower health literacy, as well as cultural factors, are determinants for non-participation in genetic counseling (Baars et al. 2017). Sharing information with the physician about family history in relation to cancer, is an important factor in the referral process. Patients with a lower social economic status, a lower educational background or a migrant background, experience greater verbal passivity and difficulties in presenting health related information to their physician (Cooper et al. 2003).

Differences in cancer incidence

Cancer incidence may vary in the population and therefore result in different participation rates in cancer genetic counseling. Migrants and people of low socioeconomic status (SES) share certain cancer risks, like the lower risks for colon, skin, breast and prostate cancer (Aarts et al. 2010; Arnold et al. 2010). Non-western migrants exhibit a higher burden of infection-related tumours (Arnold et al. 2010; Dutch Cancer Society 2006; Visser et al. 2007). The difference in cancer incidence in the population is complex, because higher cancer awareness and participation in cancer screening programs, might have contributed to a higher incidence for certain types of cancer, like breast cancer. This is usually promoted more by patients of high SES (Aarts et al. 2010). Recent research from Welch et al (2017) confirms that cancer screening is one area in which overutilization can result in over diagnosis, particularly for cancers for which the reported incidence is sensitive to early screening programs (Welch et al. 2017). Reported higher incidence does not seem to lead to a parallel increase in prevalence, as shown by the database from the National Institute of Public Health and the Environment (RIVM). This database showed no significant difference in prevalence of cancer between the lowest and highest educated people (National Institute for Public Health and the Environment 2012).

Cancer genetic counseling

In order to gain more insight in factors associated with referral to genetic counseling, we conducted an observational study in 2007 (van Riel et al. 2012). That study showed that, compared to the general population, more highly educated counselees and less migrant counselees were seen in cancer genetic counseling practice. Since 2007, more information has become available about hereditary cancer and referral for cancer genetic counseling. For physicians, guidelines have been updated and published about criteria for referral (Balmana et al. 2013; Berliner et al. 2013; Giardiello et al. 2014). Also additional value of interventions, such as an online referral test, an interactive web-based training and an electronic referral form, has been reported (Bell et al. 2015; Dekker et al. 2014; Petzel et al. 2014).

For the general public, attention to this subject has been drawn in the media, e.g. by the release of a public statement from actress Angelina Jolie (Evans et al. 2014; Roberts et al. 2017).

Over the years, for both physicians as the general population, awareness about cancer genetic counseling has increased. This is expected to be reflected in a cancer genetic counseling population which is more comparable to the general population when it comes to educational level and migrant status. To study this expectation, we conducted a study, eight years after our previous study, with the aim to determine the present-day educational level and migrant status of counselees referred to cancer genetic counseling and to investigate possible differences with our 2007 data.

METHODS

Participants

Participants were newly referred counselees for cancer genetic counseling from October 2014 to April 2015. Similar to our previous study (van Riel et al. 2012), counselees were seen by a clinical geneticist or genetic counselor from the department of Genetics of the University Medical Center Utrecht at either the university main site or one of the nine community hospitals in the region.

Study design and data collection

For each new counselee a checklist was filled in by the counselor during the first consultation. In this checklist, several items were scored: general characteristics of the counselee and the consultation, eligibility for genetic testing, educational level, country of birth of counselee and his/her parents (see Electronic Supplementary Material).

Educational level was determined by the Dutch Standard Classification of Education (Statistics Netherlands 2008) and the international classification of the UNESCO (Unesco Institute for Statistics 2011): low educational level: (pre-)primary education or first stage of

basic education; intermediate-1 educational level: lower secondary or second stage of basic education; intermediate-2 educational level: (upper) secondary education; high educational level: tertiary education. Migrant status of the counselee was determined according to the definition of Statistics Netherlands (Statistics Netherlands 2008). According to this definition, a counselee is a migrant when at least one of the parents is born outside of The Netherlands. Furthermore, a distinction can be made between Western migrants (at least one parent born outside The Netherlands, but in Europe, North America, Australia, New Zealand, Indonesia and Japan) and non-Western migrants (at least one parent born in Turkey and countries in Africa, Latin America and Asian countries). The classifications of educational level and migrant status of Statistics Netherlands was chosen to allow comparison with data about the general population of The Netherlands. Also, these same classifications were used in our earlier study (van Riel et al. 2012), so comparison with the data of the current study with the situation in 2007 is possible.

Eligibility for genetic testing was determined for the counselee or for an affected family member of the counselee based on family history and/or (if available at initial consultation) medical records, according to national guidelines for different tumor syndromes used in daily practice.

The subgroup 'other' in eligibility for genetic testing contains several reasons, e.g. eligibility can be determined after receiving the medical records of the counselee and/or family members, which are not always present at first consultation. Also in initiating discussion of family history, a category 'other' exist. This category contains initiating discussion of family history by a family member, by a family letter (a letter in case a mutation in a cancer gene is detected, intended to share with family), by the counselee and physician together or by the physician of a family member.

Statistical analysis

All data were entered in SPSS Version 21.0.0. Descriptive statistics were used to describe counselee characteristics, for university hospital and community hospital separately and for both clinics combined. Chi-square tests were used to compare the collected data to the data of the general population in The Netherlands (Statistics Netherlands 2014a; Statistics Netherlands 2014b).

RESULTS

General characteristics

In total 731 counselees were included. General characteristics, like clinical setting of the consultation, gender and personal cancer history of the counselee and eligibility for

genetic testing are shown in table 1. There were more female counselees compared to male counselees, as more than half (56%) of the affected counselees had breast cancer. About half of all counselees were seen in the university hospital (55%), the other 45% were seen in community hospitals. This is the same distribution as reported earlier in our study in 2007 (van Riel et al. 2012).

When we compare the referral between university hospital and community hospitals, we found that more counselees affected with colon cancer were seen in community hospitals, which explains the higher eligibility for microsatellite instability testing/ immune histochemistry (MSI/IHC) of counselees seen in community hospitals. In the university hospital, more counselees were seen for predictive testing for a known mutation. In initiating discussion of family history, we found discussion started by a family member in 58%, by a family letter in 23%, by the counselee and physician together in 14% and by the physician of a family member in 5%. In community hospitals, the physician more often initiated discussion of family history and less often this discussion is initiated by 'others' (e.g. a family member or via a family letter).

Educational level

When classified according to the International Standard Classification of Education (Unesco Institute for Statistics 2011), about 40% of the counselees seen for cancer genetic counseling had a high educational level. When compared for clinical setting, more counselees with an intermediate-1 and intermediate-2 level of education were seen in community hospitals and more highly educated counselees where seen in the university hospital. In comparison with the Dutch population (table 2), less counselees with a lower and intermediate-2 educational level and more highly educated counselees were seen in cancer genetic counseling. No significant difference was found in the educational level of counselees in 2007 and in 2014/2015 (van Riel et al. 2012) (table 3).

Migrant status

The majority of counselees seen for cancer genetic counseling were Dutch natives (89%). There is a trend for less migrants seen in the community hospitals ($p=0.05$). When migrants of Western and non-Western origin were compared, a significantly lower percentage of non-Western migrants is seen in community hospitals than in the university hospital. Furthermore, there were less migrants seen in cancer genetic counseling compared to the general population (table 2) ($p<0.001$). We found no significant difference in frequency of migrants referred for cancer genetic counseling 2014/2015 and 2007 (van Riel et al. 2012), (table 3).

Table 1. General characteristics of 731 counselees requesting cancer genetic counseling

variable		both clinics combined % (n=731)	university hospital % (n=403)a	community hospitals % (n=328)a	p-value
total		100.0	55.1	44.9	
gender	male	25.6 (187)	26.6 (107)	24.4 (80)	n.s.
	female	74.4 (544)	73.4 (296)	75.6 (248)	
personal cancer history	affected	50.8 (371)	46.7 (188)	55.8 (183)	0.014*
	unaffected	49.2 (360)	53.3 (215)	44.2 (145)	
affected with (n=371)	breast cancer	56.3 (209)	60.6 (114)	51.9 (95)	n.s.
	ovarian cancer	4.0 (15)	2.7 (5)	5.5 (10)	n.s.
	colon cancer	16.4 (61)	10.1 (19)	23.0 (42)	0.001*
	endometrial cancer	0.8 (3)	1.6 (3)	0 (0)	n.s.
	melanoma	2.7 (10)	4.3 (8)	1.1 (2)	n.s.
	polyposis	9.2 (34)	9.6 (18)	8.7 (16)	n.s.
	≥2 kinds of cancer	5.1 (19)	5.3 (10)	4.9 (9)	n.s.
	other[b]	5.4 (20)	5.9 (11)	4.9 (9)	n.s.
eligibility for genetic testing in counselee or relative	diagnostic DNA testing	38.2 (279)	36.0 (145)	40.9 (134)	n.s.
	MSI/IHC[c]	11.6 (85)	9.4 (38)	14.3 (47)	0.040*
	predictive testing[d]	22.0 (161)	29.5 (119)	12.8 (42)	0.000*
	did not meet criteria for testing	10.4 (76)	8.7 (35)	12.5 (41)	n.s.
	other	17.8 (130)	16.4 (66)	19.5 (64)	n.s.
initiator discussing family history (n=707)	counselee	35.6 (252)	38.3 (148)	32.4 (104)	n.s.
	physician	48.4 (342)	42.2 (163)	55.8 (179)	0.000*
	other	16.0 (113)	19.4 (75)	11.8 (38)	0.006*
educational level[e] (n=714)	low	5.0 (36)	4.1 (16)	6.2 (20)	n.s.
	intermediate-1	21.6 (154)	18.7 (73)	25.1 (81)	0.038*
	intermediate-2	35.2 (251)	31.7 (124)	39.3 (127)	0.034*
	high	38.2 (273)	45.5 (178)	29.4 (95)	0.000*
migrant status[f] (n=723)	Dutch native migrant	88.7 (641) 11.3 (82)	86.6 (342) 13.4 (53)	91.2 (299) 8.8 (29)	0.053*
	migrant, western migrant, non-western	6.7 (49) 4.6 (33)	7.1 (28) 6.3 (25)	6.4 (21) 2.4 (8)	n.s. 0.013*

** A two-sided p-value of <0.05 is considered significant*

[a] Data calculated for clinical setting (i.e. within each column)

[b] Other cancer: parathyroid adenoma, angiolipoma, carcinoid, brain tumor, hyperparathyroidism, pituitary tumor, leiomyomatosis, leukemia, neurofibroma, kidney cancer, pancreatic cancer, prostate cancer, sarcoma, sebaceoma, esophageal cancer, testis carcinoma.

[c] MSI/IHC: microsatellite instability testing / immunohistochemistry for mismatch repair deficiency

[d] Predictive testing: genetic testing for a mutation which is already known in the family of the counselee

[e] Low: (pre-)primary education or first stage of basic education; Intermediate-1: lower secondary or second stage of basic education; Intermediate-2: (upper) secondary education; High: tertiary education

[f] Dutch native: both parents are born in The Netherlands; Migrant: at least one of the parents is born outside The Netherlands. Western Migrant: at least one parent born outside The Netherlands, but in Europe, North America, Australia, New Zealand, Indonesia and Japan; Non-Western Migrant: at least one parent born in Turkey and countries in Africa, Latin America and Asian countries.

Table 2. Educational level and migrant status of counselees in cancer genetic counseling in comparison to the general population in The Netherlands

	this study (2014/2015)		general population (2014)		
	%	(n)	%	(n)	p-value
educational level					
- low	5.0	(36)	9.8	(1,229,000)	<0.01
- intermediate-1	21.6	(154)	21.0	(2,625000)	0.7033
- intermediate-2	35.2	(251)	40.7	(508,900)	<0.01
- high	38.2	(273)	28.5	(3,564,000)	<0.01
migrant status					
Dutch native	88.7	(641)	78.6	(13,234,545)	<0.001
migrant	11.3	(82)	21.4	(3,594,744)	
- western	6.8	(49)	9.5	(1,597,160)	
- non-western	4.6	(33)	11.9	(1,997,584)	

Table 3. Educational level and migrant status of counselees in cancer genetic counseling in comparison to the study in 2007 (Van Riel et al. 2012).

	this study (2014/2015)		study data (2007)		
	%	(n)	%	(n)	p-value
educational level					
- low	5.0	(36)	4.0	(16)	0.4340
- intermediate-1	21.6	(154)	26,3	(105)	0.0723
- intermediate-2	35.2	(251)	33,3	(133)	0.5400
- high	38.2	(273)	36,3	(145)	0.5314
migrant status					
Dutch native	88.7	(641)	90.6	(368)	0.2998
migrant	11.3	(82)	9.4	(38)	

DISCUSSION

Our findings suggest that patients' migrant status and educational background seem to matter in the referral to cancer genetic counseling. In 2007, we found an underrepresentation in cancer genetic counseling of migrant patients and patients with a low educational background (van Riel et al. 2012). The results of the current study show that this under-representation has not changed since then. This differential access to cancer genetic counseling may lead to treatment and outcome disparities in cancer care. The differences in educational level and migrant status, seen between counselees in the university hospital versus counselees seen in the community hospital, must be taken into account. However, definite conclusions cannot be drawn from these data because of potential differences in the patient population at the different locations. Socio demographic characteristics may have impact on patients' communicative behavior (eg asking questions or expressing concerns) as well on physicians' behavior (discussing referral possibility) (Baars et al. 2017) .

In the last years more information about cancer genetics and genetic counseling has become accessible to the general public. From popular magazines, there is an increasing focus on hereditary cancer and referral for DNA-testing. This may affect people's awareness for cancer genetic counseling and may even contribute to more patient request. As we know, patients' initiative is important in the referral process (Brandt et al. 2008; Wideroff et al. 2003). Asking questions about genetic testing increases the likelihood of being referred for genetic counseling (Al-Habsi et al. 2008; Klitzman et al. 2013). This might be an explanation for the relatively low attendance of migrant patients and patients with a lower educational background. Asking questions is associated with someone's health literacy skills (Katz et al. 2007). Health literacy skills reflect the ability to access, understand, appraise and use health-related information in various domains (Sorensen et al. 2012) and is associated with lower patient activation (Smith et al. 2013). People with a lower socio-economic position, a lower educational level and a lower subjective social status, are known to have lower health literacy skills than those with a high socio-economic position (Heide van der et al. 2013). Also many migrant patients have lower health literacy skills (Fransen et al. 2013). This might result in low awareness or understanding of familial cancer. In combination with socio-cultural variations in beliefs about cancer, this may affect patient-doctor communication, as well as referral to cancer genetic counseling (Baars et al. 2017). Given the fact that the majority of counselees seem to initiate referral to genetic counseling themselves (Brandt et al. 2008; van Riel et al. 2012), lower health literacy and corresponding lower patient activation, might contribute to the lower referral rate in migrant patients and in patients with a lower level of education. Physicians will have to adapt their communication to this group of patients in order to allow effective communication and get a higher referral rate. Recent research (Kurian et al. 2017) emphasizes the importance of oncologists' behavior in the genetic testing process. Improving their communication skills, risk estimation and optimizing triage to genetic counselors have priority.

Lower educated counselees may have other needs for genetic care than higher educated counselees (Hayat et al. 2012), which argues for a more personalized approach in both the referral process and in the genetic counseling itself. Culture-sensitive interventions can ameliorate referral to cancer genetic counseling (Hall et al. 2006). Our study points to room for improvement in referring vulnerable groups of patients. Since the outcome of cancer genetic counseling can give reasons to choose another treatment procedure, this is even more important (Christinat et al. 2013; Glenn et al. 2012; Wevers et al. 2012). Limited health literacy is likely to pose a particular challenge to cancer genetic counseling for counselees with a lower education or a migrant background. Future studies can explore how physicians should assess patients' need and skills and which communication strategies are effective.

Limitations

Our findings cannot be generalized as the study was conducted in one single clinical genetic center. However, we included a rather large number of counselees (over 700 consecutive counselees seen for cancer genetic counseling) who were seen in several hospitals in the central region of the Netherlands. Due to the study design, we don't have data about counselees who declined referral for cancer genetic counseling. These possible decliners may have had a different educational background. Also, we didn't measure the level of health literacy of the patients, but considered educational background as a proxy (Heide van der et al. 2013; Martin et al. 2009). Numerous studies show the importance of patient request or initiative in the referral process. However, in our study we found no association between initiative of the counselee and participation in cancer genetic counseling. That's probably because of bias in the scoring procedure. Although we asked who took the initiative for referral, we realize that this outcome is fairly unreliable. It's not always clear who took the initiative and sometimes the respondents didn't even remember. The observed difference in referral between university and community hospitals might be influenced by the approach of our genetic clinic: when a pathogenic mutation is identified in the index case he/she receives a family letter to inform family members. With this letter, family members can directly contact our department at the university hospital, and are more often invited for a consultation at this location. Furthermore, more information about characteristics of the referring physicians and their practice might lead to a better clarification of the differences seen between consultations in the different clinical settings. As cancer incidence, as well as demographics of the Dutch population, vary over years, this may influence the referral for cancer genetic counseling. In our study we did not standardize for these differences. Related to migrant status and socioeconomic inequalities, a variety of studies from Europe has shown that disparities in the burden of cancer exist (Arnold et al. 2010). In further research we must take these differences into account.

To conclude: the results in this study are similar to the results in 2007. Lower participation in cancer genetic counseling by migrant patients and patients with a lower educational background is still a cause for concern. Additional research on interventions how to improve referral for these patients is urgently desired.

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Compliance with ethical standards

Conflict of interest

The authors declare no conflict of interest.

Ethical approval

All procedures followed were in accordance with the ethical standards of the responsible clinic. For this study, no ethical approval was required, because most items of the checklist are discussed routinely in regular consultations. We didn't ask the counselees to perform any action, like filling out questionnaires.

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SUPPLEMENTARY MATERIAL: CHECKLIST

Clinical setting: ❑ university hospital ❑ community hospital	Date of consultation:
Research number:	

Disease status: ❑ unaffected ❑ affected: → ❑ breast cancer
❑ other: ……

Time-span for genetic testing: ❑ regular
❑ rapid genetic testing

Reason for referral:
❑ hereditary breast cancer
❑ Lynch syndrome
❑ other tumor predisposition syndrome: …….

DNA-testing:
❑ diagnostic DNA-testing
❑ predictive testing
❑ microsatellite instability / immunohistochemistry
❑ not eligible
❑ other: …….

Highest completed education by counselee*:
❑ no education
❑ primary education
❑ lower secondary education
❑ preparing for vocational education
❑ general and vocational programs preparing for tertiary education
❑ higher secondary education
❑ tertiary education

*Low = Low: (pre-)primary education or first stage of basic education;
Intermediate-1: lower secondary or second stage of basic education
Intermediate-2: (upper) secondary education
High: tertiary education

Who initiated discussion of family history?
❑ Counselee
❑ Physician
❑ Other:……..

In which country are counselee and his/her parents born?

Counselee:	❑ The Netherlands	❑ Other: …….
Father:	❑ The Netherlands	❑ Other: …….
Mother:	❑ The Netherlands	❑ Other: …….

PART II

Development of a health literacy training program and a plain-language guide

CHAPTER 3

3

Systematic development of a training program for healthcare professionals to improve communication about breast cancer genetic counseling with low health literate patients

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Abstract

There is a disproportionate underuse of genetic testing in breast cancer patients from lower education or migrant background. Within these groups, communication about referral to genetic counseling appears challenging due to limited health literacy and cultural barriers. Our aim was to develop and evaluate a training program for healthcare professionals (breast surgeons and specialized nurses), to increase effective communication. We systematically developed a blended training program based on patients' and healthcare professionals' needs and preferences. Prior to the training, we assessed awareness, knowledge and self-efficacy of healthcare professionals. Acceptability and usefulness of the training program were assessed directly after the training. Healthcare professionals (n=65) from 17 hospitals showed moderate to high awareness and knowledge about the prevalence and impact of limited health literacy. They were aware of cultural factors that influence communication. However, they did not feel confident in recognizing limited health literacy and their self-efficacy to communicate effectively with these patients was low. The training program was rated as acceptable and useful. Healthcare professionals lack confidence to effectively communicate with patients with limited health literacy or migrant background. The training program offers opportunities to improve communication about referral to breast cancer genetic counseling.

Keywords: blended training program; breast cancer genetic counseling; communication skills; disparities; health literacy; migrant status; referral.

INTRODUCTION

Referral to genetic counseling for breast cancer patients at risk of carrying a mutation is crucial and should preferably be offered early after diagnosis to guide treatment decisions. Breast cancer patients with a BRCA 1/2 gene mutation can decide whether or not to opt for bilateral mastectomy as primary surgery and also the chemotherapeutic approach for these patients can be different[1-3]. In addition, an abnormal test result may have important implications for cancer prevention strategies for patients and their (healthy) family members, including future generations. Although genetic counseling is clinically relevant for all eligible high-risk patients with breast cancer, there's still a disproportionate underuse of it in patients with a lower educational background and in migrant patients[4-7]. These patients seem to have poorer access to cancer-related genetic counseling[7,8]. Patients need to understand the benefits, limitations and risks of genetic testing, value this information, communicate about it properly with healthcare professionals and family members and make an informed decision regarding the possible consequences of a genetic test result. This requires adequate health literacy, which is generally defined as a persons' ability to access, understand, evaluate and use health-related information and is recognized as a critical factor affecting communication in cancer care[9]. Research shows that poor awareness of family history, inaccurate risk perception and a lack of awareness of genetic services contribute to patients' misunderstanding of genetic services[10,11]. Besides, patients with limited health literacy also show a lower preference for active participation in decision-making about genetic testing[12] and in taking initiative for referral to genetic counseling. For migrant breast cancer patients, language difficulties and limited health literacy, as well as cultural factors, are determinants for non-participation in genetic counseling[13].

However, physicians also contribute to these disparities in access to breast cancer genetic counseling in the way they communicate. Provider recommendation is a first step towards uptake of genetic counseling[14] but referral is not always adequately discussed with patients with limited health literacy[15-18]. Women attribute their low levels of awareness of genetic testing to a lack of physicians' recommendation for referral, which they also noted as their primary reason for not receiving testing[17,19]. Baars et al. showed that a major cause for the low participation rate in cancer genetic counseling lies within the referral process. Although referral guidelines are sufficiently available and known by physicians, they do not always act in concordance with these guidelines[20,21]. Gaps in effective communication are widely recognized as a major contributor to health disparities[22] also in the genetic testing[17]. Employing effective communication techniques for healthcare professionals is an important intervention to reduce health disparities related to limited health literacy[23]. However, implementation of a training program for healthcare professionals is a complex process. Successful adoption is only possible if healthcare professionals themselves deem it useful[24] and when they are involved during the development of the program[25]. The aim of the present

study was to develop a training program for healthcare professionals (breast surgeons and specialized nurses) to communicate effectively about referral to breast cancer genetic counseling with patients with limited health literacy or a migrant background. Specific objectives were: 1. to develop a training program based on the needs and preferences of healthcare professionals and patients, 2. to assess knowledge, awareness and self-efficacy in communication with patients with limited health literacy or a migrant background, and 3. to gain insight in the usefulness and acceptability of the training program from healthcare professionals' perspective.

MATERIALS & METHODS

Development of the training program

We systematically developed a blended training program (Erfo4all), consisting of an online module and a group training, based on healthcare professionals' and patients' needs and preferences. The intervention mapping (IM) approach[26] a protocol for developing theory- and evidence-based health education programs was used as a helpful guideline (Supplementary table 1).

Based on (a) the needs and preferences of healthcare professionals and patients and insights from our previous studies and (b) a matrix of change performances and objectives, we (c) made deliberate choices regarding the design and content of the training program and (d) pilot-implemented the program in clinical practice. Each step in the development process is described in detail below.

Assess needs and preferences of healthcare professionals and patients

We conducted a group interview with breast surgeons and specialized nurses, who are the main referrers to genetic counseling for patients with breast cancer[8,27] to assess their preferences regarding content and design of a training program and to gain insight into conditions to participate. They were recruited from different breast cancer teams from different hospitals in Western Netherlands. For the content of the training program from patients' perspective we elaborated on findings from our previous study on participation determinants and perspectives of (migrant) patients and healthcare professionals in breast cancer genetic counseling[13,20]. In the present study we conducted in-depth interviews with three patients to deepen the relevance of our findings. Patients were asked to share their experience with breast cancer genetic counseling and state barriers, needs and preferences for communication with their surgeon or specialized nurse. They were recruited in collaboration with Mammarosa, an organization that provides information about breast cancer for migrant patients and patients with a low level of literacy. The patients were able to speak Dutch and had personal experience with breast cancer care and cancer genetic counseling. The

interviews with healthcare professionals and patients were audio-recorded and transcribed verbatim to increase validity. Analyses were done by two authors, working independently. Seven healthcare professionals (three breast surgeons, one medical oncologist and three specialized nurses) participated in the group interview. The group interviews indicated that healthcare professionals experience difficulties in recognizing patients with limited health literacy in daily practice and a tendency to overestimate the health literacy skills of their patients. They also indicated a need for more information and for tools to communicate effectively about referral to breast cancer genetic counseling with patients with limited health literacy or a migrant background, Healthcare professionals had a clear preference for a blended learning intervention, consisting of an online module followed by a multidisciplinary group training of limited duration to enhance their skills in tailoring communication about genetic testing to patients with limited health literacy. Patients noted communication with the physician or specialized nurse as the most important factor influencing referral. They stated that the use of plain language, non-medical jargon and tailored information are very important in communication about breast cancer genetic counseling. Patients mentioned various difficulties with taking initiative for referral to breast cancer genetic counseling. They experienced insufficient knowledge and skills to discuss referral possibility with their physician when they were diagnosed with breast cancer. Asking questions was considered to be difficult for most migrant patients. According to patients, taking into account social and cultural beliefs about cancer and also the use of a professional interpreter contribute to more effective communication about referral to breast cancer genetic counseling.

Matrix of change performances and objectives

Based on healthcare professionals' training preferences and input from patients' perspective on the content of the training we specified performance and change objectives in a matrix of change (Supplementary table 2).

We then selected various practical strategies from literature to improve communication with limited health literate patients, such as information transfer to enhance knowledge and awareness about the problem of limited health literacy[28] and the Teach-back method to identify patients with limited health literacy[23,29]. Role-play[30] was selected as a strategy to acquire required communication skills and to practice the use of plain language and the Teach-back method. To further enhance health professionals' ability to communicate in an effective manner with patients with a migrant background, we selected strategies to enhance cultural competences[31].

Design and content of the training program

In the next step, the practical strategies were incorporated into a blended training program (Erfo4all), consisting of two successive parts: an online module (18 minutes) and a group training (2 hours). The online module focused on knowledge acquisition, while in the group

training practicing skills were most important. An online module offers opportunities to increase knowledge, but it is likely not sufficient for behavior change. Integrating an online training with traditional face-to-face training gives the opportunity to increase knowledge as well as practical skills. The background information in the training program was based on the reports on health literacy from the national institute for health services research in the Netherlands[32,33]. We used video-recordings from the Dutch Reading and Writing foundation to include patients' perspective in the background information. In these video-recordings low literate people shared their experience with being low literate, talked about their shame and explained how they tried to hide their problem in real life. Information about the prevalence of low literacy and limited health literacy in the Netherlands, the relevance of health literacy in understanding and appraising information from healthcare professionals as well as the way health literacy relates to socio-economic and demographic characteristics were incorporated in the online module. Also specific attention was given to communication with patients with a migrant background, including the impact of a language barrier and cultural factors on communication with healthcare professionals. The training methods were developed in collaboration with Pharos (Dutch Centre of Expertise on Health Disparities). We made use of their group training on effective communication with patients with limited health literacy or a migrant background and adapted it to the context of clinical genetics and breast cancer genetic counseling. Roleplay and the teach-back method already were key elements in their training and were further refined to reach our performance objectives. In cooperation with clinical geneticists, we added real-life cases, with a focus on migrant and non-migrant patients, in relation to cancer genetic counseling.

Pilot-implementation of the training program

We pilot-implemented the Erfo4all training program in 17 hospitals in three regions in the Netherlands. Healthcare professionals from these hospitals refer breast cancer patients for genetic counseling to clinical geneticists of one of three academic centers. Together with clinical geneticists from these three contributing academic centers, we developed a detailed plan on recruitment for breast surgeons and specialized nurses in referring hospitals, including instructions for contact persons to motivate colleagues to participate in the training. The Center for Research and Development of Education from the University Medical Center Utrecht created private accounts for the participants of the training program, which gave them access to a questionnaire and the online module. Accreditation by the Dutch Association for Surgery and the Dutch Professional Nurse Practitioner Organization was an incentive for participation.

Assessment of knowledge, awareness and self-efficacy

Before the training, we assessed healthcare professionals' knowledge, awareness and self-efficacy regarding communication with patients with limited health literacy or a migrant

background using an online questionnaire. Knowledge was assessed with five multiple choice questions focusing on:

- prevalence of low literate adults in the Netherlands. Answers ranging from (A) to (D).
- limited (health) literacy in relation to people with a migrant background. Answers ranging from (A) to (C).
- prevalence of adults with limited health literacy in the Netherlands. Answers ranging from (A) to (D).
- level of education in relation to the level of health literacy. Answers ranging from (A) to (C).
- use of a professional interpreter (self- reported). Answers (yes) or (no).

Each item was rated as correct (1) or wrong (0) and a total knowledge score was computed as the number of correct answers.

Awareness was assessed by three items on:

- prevalence and impact of health literacy in the Netherlands
- impact of health literacy on medical communication
- importance to take into account cultural factors in communication with patients with a migrant background.

Each item was scored on a 5 point Likert scale ranging from (1) low, to (5) very high.

Self-efficacy was assessed by five statements on having confidence in:

- recognizing limited health literacy in patients
- communicating effectively about breast cancer genetic counseling with patients with limited health literacy
- understanding which customs and habits from patients with a migrant background might influence communication
- coping with cultural factors in communication with patients with a migrant background
- coping with a language barrier

Each item was scored on a 5 point Likert scale ranging from (1) totally disagree to (5) totally agree. Descriptive statistics were used to describe baseline characteristics and outcome variables using SPSS version 24.0 (SPSS, Chicago, IL). Data was used as baseline measurement for our study on effectiveness of the training program.

Test training program on acceptability and usability

After completing the questionnaire healthcare professionals got access to the online module and within two weeks they were invited for the group training on location. Each healthcare professional completed a paper-and-pencil evaluation survey after completion of the training program. The evaluation survey contained five questions assessing acceptance of the program, measured on a 5-point Likert scale ranging from (1) very good to (5) not good at all. The following items were assessed:

- design of the online module
- duration of the online module
- blended learning method
- duration of the group training
- time schedule of the group training

In addition, participants were asked to rate the usefulness of the training program measured on a five point Likert scale, ranging from (1) very useful to (5) not useful at all. The following items were assessed:

- online module
- group training on location
- training elements
  - recognizing low literacy/limited health literacy (teach-back method)
  - general advice to communicate in plain language
  - obtaining family history
  - cultural sensitive communication
  - specific advice to communicate in plain language about (referral to) breast cancer genetic counseling
  - practicing real life cases (role-play)

Finally the quality of the module and the group training, as well as competence of the trainer and the training actress were rated on a scale from 1 (low) - 10 (high).

RESULTS

Response and characteristics of participants of the training

A total of 73 healthcare professionals were included in the training program. The online questionnaire was completed by 65 healthcare professionals from 17 hospitals. Table 1 shows an overview of background characteristics.

Awareness, knowledge and self-efficacy healthcare professionals

Prior to the training, the majority of healthcare professionals showed a moderate to high score on awareness about the prevalence and impact of health literacy in the Netherlands, as well as on the impact of limited health literacy on medical communication (table 4). They were highly aware of the importance to take into account cultural factors in the communication with patients with a migrant background and 46% reported to deploy a professional interpreter in communication with patients with a language barrier.

Table 1. *Background characteristics of healthcare professionals (n=65).*

variable		N	%	Mean (sd)
sex	male	12	18.5%	
	female	53	81.5%	
discipline	breast surgeon	21	32.3%	
	specialized nurse	38	58.5%	
	medical oncologist	1	1.5%	
	physician assistant	2	3.1%	
	other	3	4.6%	
age				45.7 (8.5)
work experience in breast cancer care (in years)				10.9 (7.0)
clinical setting	university hospital	6	9.2%	
	community hospital	59	90.8%	

Knowledge about prevalence of limited (health) literacy in the Netherlands was moderate (mean accurate knowledge score was 2.48, sd.98) and most healthcare professionals knew which factors are related to limited health literacy. Self-efficacy in communication with patients with limited health literacy or a migrant background however, was low. Healthcare professionals reported to frequently encounter challenges in recognizing limited health literacy in patients, to communicate effectively about breast cancer genetic counseling and to cope with cultural factors in the communication with patients with a migrant background.

Acceptability and usefulness of the training program

The training program was evaluated positively by the healthcare professional. They reported a high degree of acceptance with the blended learning method; the combination of an online module and a group training on location was considered useful and time-efficient. They were satisfied with the duration of the training, both the module and the group training, as well as with the design of the module. Furthermore, the healthcare professionals appreciated the trainer and the training actress, the average score for the training actress was 9.3 and for the trainer 9.0 on a scale from 1 to 10. Figure 1 shows perceived usefulness of training elements. Training elements with a high score included recognizing low literacy/ limited health literacy, general advice on how to communicate in plain language, assessing family history, cultural sensitive communication, communication about breast cancer genetic counseling in plain language and practicing with real-life cases. Most healthcare professionals would recommend the training to their colleagues. Overall, the participants' evaluation suggests that the training program was well accepted.

Table 2. Awareness, knowledge and self-efficacy of healthcare professionals prior to the training.

	n	%
Awareness		
Awareness of prevalence and impact of health literacy		
• low	-	-
• barely	4	6.2%
• reasonably	49	75.4%
• high	10	15.4%
• very high	2	3.1%
Awareness of impact health literacy on communication		
• low	-	-
• barely	3	4.6%
• reasonably	33	50.8%
• high	27	41.5%
• very high	2	3.1%
Awareness of the importance assess cultural factors		
• low	1	1.5%
• barely	1	1.5%
• reasonably	10	15.4%
• high	42	64.6%
• very high	11	16.9%
Knowledge		
Prevalence of illiteracy in the Netherlands		
• correct answer	46	70.8%
• wrong answer	19	29.2%
Limited (health) literacy and a migrant background		
• correct answer	38	58,5%
• wrong answer	27	41,5%
Prevalence adults with limited health literacy		
• correct answer	20	30.8%
• wrong answer	35	55.4%
Level of education related to level of health literacy		
• correct answer	48	73.8%
• wrong answer	17	26.2%
Use of professional interpreter		
• correct answer	58	89.2%
• wrong answer	7	10.8%
Sum scores / Total knowledge (Mean 2.48, sd .98)		
• 1	2	3.1%
• 2	14	21.5%
• 3	21	32.3%
• 4	23	35.4%
• 5	5	7.7%

	n	%
Self-efficacy		
Having confidence in understanding which customs and habits from patients with a migrant background might influence communication		
• totally disagree	2	3.1%
• disagree	28	43.1%
• not agree/not disagree	27	41.5%
• agree	7	10.8%
• totally agree	1	1.5%
Having confidence in recognizing limited health literacy		
• totally disagree	-	-
• disagree	9	13.8%
• not agree/not disagree	30	46.2%
• agree	26	40.0%
• totally agree	-	-
Having confidence in communicating effectively about breast cancer genetic testing with patients with limited health literacy		
• totally disagree	-	-
• disagree	17	26.2%
• not agree/not disagree	36	55.4%
• agree	12	18.5%
• totally agree	-	-
Having confidence in coping with cultural factors		
• totally disagree	-	-
• disagree	16	24.6%
• not agree/not disagree	29	44.6%
• agree	20	30.8%
• totally agree	-	-
Having confidence in coping with language barriers		
• totally disagree	-	-
• disagree	9	13.8%
• not agree/not disagree	30	46.2%
• agree	26	40.0%
• totally agree	-	-

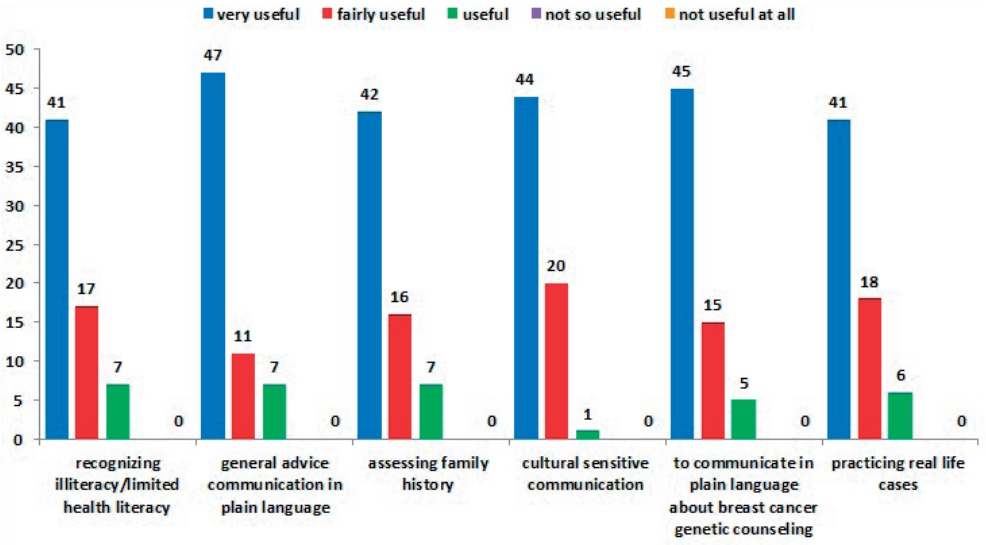

Fig. 1 *Perceived usefulness of training elements.*

DISCUSSION

In this paper we described the systematic development, pilot-implementation and acceptability of a blended training program for breast surgeons and specialized nurses to improve communication about referral to breast cancer genetic counseling. The content and format of the program was based on their training needs and preferences and tailored to patients' perspective. Upon the training, healthcare professionals were aware of the problem of limited health literacy and reported to have knowledge about prevalence of low literacy, limited health literacy and the main factors associated with health literacy. However, they didn't feel competent to recognize limited health literacy and to communicate effectively with these groups of patients. The training program was evaluated as acceptable on method, design and duration, and participants rated the digital module, group training and training elements as useful.

The needs and preferences that we obtained during the development of the training program, indicated that healthcare professionals experience difficulties in recognizing limited health literacy in patients and are in need of techniques to communicate effectively about referral to breast cancer genetic counseling with patients with limited health literacy or a migrant background. This need for training has been reported by others as well[34-36]. Coelho (2018), for example, found that 84% of the healthcare professionals would like more training on health literacy, including assessment tools and techniques to manage limited health literacy.[34] An unexpected finding of our study was that healthcare professionals' knowledge and awareness regarding prevalence and impact of limited health literacy and

cultural factors influencing communication was generally adequate. Other studies indicated lower perceived awareness and knowledge about health literacy[34,36]. Although healthcare professionals seem generally aware of cultural differences, different studies indicate that awareness is not enough. Enhancing cultural competence, the ability to cope with cultural differences, is important to communicate effectively with patients with a migrant background[37,38]. The outcomes of the questionnaires further indicated that healthcare professionals' self-efficacy to communicate with patients with limited health literacy or a migrant background was low. Therefore, improvement of healthcare professionals' self-efficacy to communicate with patients with limited health literacy or a migrant background is important, especially because self-efficacy is related to one's competence and to future (communication) behavior. Knowledge alone is insufficient for actual behavior change. Therefore, using role play, focusing on plain language, using the teach-back method and cultural sensitive communication, are key elements in our training program. We choose for role play because this is an effective training strategy to practice and learn communication skills[39]. Other studies showed promising results regarding the use and effectiveness of the teach-back method in communication with patients with limited health literacy[40] and cultural sensitivity training for improved understanding of cultural factors and the ability to communicate with patients with a migrant background[31]. Participants in our study were very positive about the acceptability and usability of the training, this is important for adoption and successful implementation of the program. Implementation effectiveness is critical for transporting interventions to daily practice[41]. Because of the high acceptance of the program and focus on enhancing skills, the Erfo4all training program seems to offer opportunities to improve communication about breast cancer genetic counseling. The setting of breast cancer genetic counseling is not unique compared to genetic counseling for other types of cancer or even genetic disorders. In general, limited health literacy is associated with lower genomic related knowledge and it affects patients' understanding of print and oral communications about genetic and genomic information, so adapting communication to patients with limited health literacy is important in different settings of genetic counseling. We think is feasible to adapt our program to these other settings. The next step in our research is to study the effectiveness of the Erfo4all training program on knowledge, awareness and self-efficacy regarding communication with patients with limited health literacy or a migrant background.

Strength and limitations

A strength of this study was the systematic approach in the development of the training. The needs and preferences of healthcare professionals and patients were used to determine the format and content of the program and to enhance a successful implementation. However, there are also some potential limitations. First, we included healthcare professionals in the training program on a voluntary base, so selection bias cannot be ruled out. Healthcare

professionals who are already more aware of the problem of limited health literacy and have a basic knowledge about the subject, may be more interested in participating in the training program. And second, we assessed awareness and self-efficacy by a self-reported instrument, so bias, like social desirability, may affect the results. Our study emphasizes the need and feasibility of a training program for healthcare professionals in the context of clinical genetics and can be used to improve communication about breast cancer genetic counseling with patients with limited health literacy or a migrant background. We are currently performing a study to find out whether this training program contributes to a higher referral rate and increased access to breast cancer genetic testing for these groups of patients. In this study we specifically developed a training program for healthcare professionals and not for patients. In future research it may be worthwhile to consider whether empowering patients (e.g., by asking questions, or by taking the initiative to discuss possible genetic causes of their breast cancer) can also contribute to effective communication about referral to breast cancer genetic counseling.

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Compliance with ethical standards

Ethical approval

All procedures followed were in accordance with the ethical standards of the responsible clinic. Participants of the focus group interviews gave permission to audio-record the interview for study purposes.

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Supplementary table 1. Steps in systematic development of the training program; intervention mapping approach

Step	Task
1. needs assessment	specify needs of healthcare professionals (breast surgeons and specialized nurses) specify needs of patients
2. definition of performance and change ojectives	establish a matrix of change
3. selection of intervention, methods and strategy	determine focus of the intervention
4. design and production of the intervention	design of the training program
5. implementation plan	develop an implementation plan to examine the effect of the training program
6. evaluation plan	describe methods for evaluation of the ntervention

Supplementary table 2. Matrix of change, performance objectives and change objectives in relation to communication about (breast) cancer genetic counseling

Performanceobjectives	**Determinants**			
	awareness	*knowledge*	*attitude*	*skills*
	change objectives			
Health professionals recognize signs of low literacy or limited health literacy in patients	Health professionals are aware of the fact that many people in the Netherlands have difficulties with reading, understanding and processing (medical) information.	Health professionals know which (group of) patients are most at risk in being low (health) literate Health professionals know which communication techniques are most appropriate in order to detect low literacy or limited health literacy.	Health professionals consider it important to recognize low literacy or limited health literacy in patients.	Health professionals can apply specific communication techniques to detect low literacy or limited health literacy in patients.
Health professionals communicate effectivelyabout breast cancer genetic counseling with patients with low literacy or limited health literacy	Health professionals are aware of the differences in referral to cancer genetic counseling between women with a low educational background or migrant patients and women with a high educational background. Health professionals are aware of their (shortcomings in) communication skills.	Health professionals know how to explain information about breast cancer genetic counseling in plain Dutch to patients with low literacy, limited health literacy or a migrant background.	Health professionals consider it important to tailor their communication and explain information about breast cancer genetic counseling in plain Dutch.	Health professionals adapt their communication to the needs and abilities of the patient and his or her family.
Health professionals adequately cope with a language barrier	Health professionals are aware of the impact of a language barrier on medical consultation.	Health professionals know why a professional interpreter is preferable to a family interpreter. Health professionals know how to access the services of a professional interpreter.	Health professionals have a positive attitude toward the use of a professional interpreter instead of family interpreter.	Health professionals make adequate use of the services of a professional interpreter.
Health professionals act in a culturally sensitive manner	Health professionals are aware of their knowledge and beliefs about people with a different cultural background.	Health professionals know how to take into account different cultural, psychological and religious aspects in their communication with patients with a migrant background and their family members.	Health professionals consider it important to take into account cultural aspects.	In their communication with patients with a migrant background, health professionals take into account cultural aspects, such as a taboo on talking about cancer.

CHAPTER 4

4

Development of a plain-language guide for discussing breast cancer genetic counseling and testing with patients with limited health literacy

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Abstract

Purpose : Due to limited health literacy and resulting ineffective communication between healthcare professionals and patients, not all eligible patients are offered breast cancer genetic counseling and testing. We aimed to develop a plain-language guide to increase effective communication about genetic counseling and testing with breast cancer patients with limited health literacy.

Methods: Together with oncological healthcare professionals we drafted a list of jargon words frequently used during (breast) cancer genetic counseling. In a focus group interview with breast cancer counselees with limited health literacy, who had received genetic counseling before, we reformulated these words in plain language. Low-literate individuals, who are not familiar with breast cancer care or genetic counseling, reflected on the draft of the guide. Completeness, acceptability and perceived usability were tested in an online questionnaire among healthcare professionals.

Result: The result is a plain-language guide for genetic counseling and testing with 33 frequently used jargon words and a reformulation of these words in plain language. Acceptability and perceived usefulness of the guide among healthcare professionals (n=58) was high.

Conclusion: The plain-language guide provides opportunities to facilitate communication about genetic counseling and testing with patients with limited health literacy and could enhance opportunities for patients to make informed decisions to participate in genetic testing. As the intention from healthcare professionals to use the plain-language guide is high, implementation of the guide in a real-life setting seems promising.

Keywords: Genetic counseling and testing, plain language, health literacy, genetic literacy, access to care.

INTRODUCTION

It is important that women at risk of carrying a mutation in a breast cancer gene are offered breast cancer genetic testing.It can help them to make decisions about their own treatment or prevention strategies and can have implications for their (healthy) family members, including future generations[1,2].

Due to limited health literacy and resulting ineffective communication between healthcare professionals and patients, not all eligible patients are offered genetic counseling and testing[3-9]. Patients' limited health literacy and their lack of experience with the healthcare system were found to be barriers, making it difficult for patients to actively engage in taking healthcare decisions[10,11] and is also associated with lower genomic related knowledge[12,13]. Given that in the Netherlands over 36% of Dutch adults have low or limited health literacy[14], a sizeable proportion of patients lack adequate understanding of medical terms. Most health literacy projects have focused on patient factors, with relatively less emphasis on the communication skills of healthcare professionals[15,16]. However, being able to correctly assess the patient's level of health literacy, is a prerequisite for effective communication. Research shows that there are significant gaps in knowledge, awareness and skills to recognize limited health literacy among nurses and physicians[17-19].

In daily practice, jargon is overused in communication with patients and is a barrier to effective medical communication, especially when health literacy is limited or the topic is complicated[20,21]. Avoiding jargon and using plain language seem promising strategies for effectively communicating health information.[22-24]. In the context of genetic counseling it was found that the greater the use of technical terms, the greater the literacy demand of a genetic counseling session[25]. Guidelines or tools for the use of plain language may be a useful addition to medical consultations[26]. Although there are a number of plain-language word replacement resources, like a plain-language medical thesaurus[27], these tools are not sufficiently tailored to the context of (cancer-)genetics. In the context of genetics, the development of the REALM-G recognizes the need to identify which patients may be in need of communication in plain language because of limited health literacy[28]. However, it can not be used as a tool to facilitate healthcare professionals to communicate effectively about breast cancer genetic counseling.

The specifics of plain language tools depend on the needs of patients so it is critical to involve them in the development process[22]. But also involving healthcare professionals as intended end-users is crucial for effective implementation. Solutions designed in this way are more likely to be acceptable to both providers and end-users[29].

We aimed to develop together with breast cancer patients with limited health literacy and low- literate individuals a plain-language guide for healthcare professionals to effectively discuss breast cancer genetic counseling and testing. The following research questions were addressed:

What are plain language synonyms for jargon words frequently used in breast cancer genetic counseling and testing according to breast cancer patients with limited health literacy and low-literate individuals?

How do intended end-users (healthcare professionals) perceive the completeness, acceptability and usefulness of a plain-language guide for genetic counseling?

METHODS

The development of the plain-language guide is part of the Erfo4all project, a project that aims to achieve equal access to breast cancer genetic counseling for all eligible patients. Within this project, we developed a blended training program for healthcare professionals, consisting of two successive parts: an online module and a group training[30]. In the group training, the teach-back method - a methodology used by healthcare professionals to check whether a patient understands what has been discussed - was used as a technique to identify patients with limited health literacy[31].

Participants

The plain-language guide was developed step by step, using an iterative two-stage design. Breast cancer patients with limited health literacy, low-literate individuals and intended end-users (breast surgeons, clinical geneticists and specialized nurses) were actively involved.

Instrumentation and procedures

Phase 1: Focus group interviews breast cancer patients with limited health literacy and low-literate individuals.

Together with breast cancer surgeons and specialized nurses (n=59) who completed the Erfo4all training program[30], a clinical geneticist and a genetic counselor from the Genetics Department of the University Medical Center Utrecht, we drew up a list of jargon words that are frequently used verbally and in writing during breast cancer genetic counseling consultations.

Subsequently, we conducted a focus group interview with breast cancer patients with a lower educational background or a limited level of health literacy and a personal experience with breast cancer genetic counseling, and reformulated these words in plain language. Input from the focus group interview was used to develop a draft of the plain-language guide.

In a second group interview with low-literate individuals with no personal experience in breast cancer genetic counseling, the first draft of the guide was evaluated. We conducted this second group interview because there is evidence that a sizeable proportion of laypersons

lack adequate understanding of several common terms used in medical consultations, do not understand phrases often used in cancer consultations and cannot be assumed to have basic medical knowledge[32,33]. Participants were asked to provide feedback relating to the comprehensibility of the preliminary version of the plain language guide thereby supported by an information letter in which the setting of breast cancer genetic counseling was outlined. Based on the feedback of low literate individuals, we refined the guide.

Participant selection

For the first focus group interview, we wanted to include patients with limited health literacy to provide input for a language guide adapted to their needs and abilities. Breast cancer patients who completed breast cancer genetic counseling at the Genetics Department of the University Medical Center Utrecht between March 2017 and October 2018, were invited. Selection of these patients was done using background data that were registered on a checklist of the Erfo4all project.We selected patients that either scored low on health literacy, or had a low educational attainment and migrant background, because these variables are known to be associated with health literacy competences[34]. Health literacy was assessed by asking patients the validated question: 'How often do you need help reading letters or information from your doctor, hospital or other health institutions?'[35]. Inclusion criteria were: no medical or social restriction for participation and able to speak Dutch. Eligible patients (n=64) received a letter in plain Dutch to inform them about the aim and the procedure of the focus group interview. Within two weeks a researcher contacted them by phone to ask if they wanted to participate. For the second group interview, low-literate adult individuals with no personal experience with breast cancer genetic counseling, recruited from the Dutch Reading & Writing Foundation were invited. They also received a letter in plain Dutch to inform them about the aim and the procedure of the group interview and an invitation to the meeting. Ethical approval for the study was waived, but in line with the declaration of Helsinki[36], we asked participants from both group interviewsto sign a consent form, certifying that the information given is confidential, that participants understood the study information and that they are aware of the fact that they can withdraw from the focus group interview any time. They also gave permission to audio-record the interview.

Data collection during focus group interviews

Patients were asked to reflect on jargon words used during routine breast cancer genetic counseling. We asked which words were unknown or difficult, which words they recognized and what they thought the meaning of these words was. Together with the patients, we rephrased difficult words concerning genetic counseling and testing in plain language until the participants were satisfied with the final formulation. During the interviews we used the teach-back method as a strategy to ensure words and explanations are understood[37,38]. In the group interview with participants from the Dutch Reading & Writing Foundation,

the guide was discussed and tested on laypersons' understanding. Both group interviews lasted 1.5 hours and were audio-recorded, so they could be listened to independently by two authors (JG and SvD) to ensure no information was missed.

Phase 2: Survey among intended end-users

In this phase we aimed to explore intended end-users' (breast cancer surgeons, specialized nurses, clinical geneticists, genetic counselors) perceptions of the plain-language guide on completeness, acceptability and usefulness. The plain-language guide and a digital questionnaire were sent to 59 healthcare professionals involved in breast cancer care in three regions in the Netherlands, who participated in the Erfo4all training program and to clinical geneticists and genetic counselors from the genetics departments in four academic centers in the Netherlands (n=47). A cover letter informed them about the aim of the study and the importance of their input. We asked healthcare professionals if they noticed any unnecessary or missing words on the preliminary list. Further, we asked about their acceptance of the reformulation in plain language, the perceived usefulness of the guide and finally their intention to use the guide in daily practice. We used an adapted version of the USE questionnaire[39] to assess the acceptability and perceived usefulness, based on a 5-point Likert scale, ranging from (1) totally disagree to (5) totally agree. Open-ended questions were used to ask about their intentions to use the plain-language guide and to ask for suggestions to refine the guide on content or design.

Statistical analyses

All data from the questionnaires were entered in SPSS Version 24.0. Categorical data, number of healthcare professionals, sex and discipline are presented in numbers and percentages. Descriptive statistics were used to present outcome measures from the questionnaires.

RESULTS

Outcomes phase 1: Feedback from breast cancer patients with limited health literacy and low-literate individuals

Response

Of the 64 patients who were invited, 11 patients and four of their partners participated in the focus group interview. Table 1 shows the background characteristics of participating patients.

All patients had a lower level of education (i.e. less than primary education, primary or lower secondary education) or were identified to have limited health literacy. Patients who did not participate explained that this was due to practical considerations, mostly involving their work schedule or transportation to the hospital. In the second session with participants

Table 1. *background characteristics of patients participating in the focus group interview*

		n
sex	male	2
	female	9
breast cancer	yes	7
	no	4
eligibility for genetic testing	diagnostic DNA testing	7
	predictive testing	3
	did not meet criteria for testing	1
educational level	low	4
	intermediate-1	7
	intermediate-2	0
	high	0
level of health literacy	low	10
	intermediate	1
	high	0
migrant background	yes	2
	no	9

from the Reading & Writing Foundation, three low literate individuals participated; one male and two females.

Reflection on jargon words and reformulation by (breast cancer) patients with limited health literacy (focus group interview 1)

Patients with limited health literacy stated that terms related to genetic testing are difficult to understand and sometimes ambiguous, *[e.g., 'hereditary or genetic predisposition, what's the difference?']*. Moreover, the difference between a gene and DNA needed clarification. Jargon words '(gene) mutation' and 'gene panel' are considered the most difficult and abstract words. A gene panel is associated with a group of individuals and not with a test that analyzes multiple genes at once for cancer-associated mutations *['I think we are in a gene panel right now']*. According to patients, it is important to be as specific as possible and to avoid abbreviations.

Reflection by low-literate individuals on the draft version of the plain-language guide (focus group interview 2)

Low-literate individuals considered most of the jargon words in the plain-language guide and in the patient information letter to be difficult. *['these are all difficult words']*. They stressed the importance of meeting the needs of patients with lay knowledge *['it's another world, we have no idea']* and stated that most of the rephrased words on the plain-language guide are acceptable and understandable. Based on the patient information letter, participants from this group interview suggested four more jargon words and the reformulation of these words, to add to the plain-language guide. Table 2 shows the primary list of jargon words,

Table 2. Reflection by patients with limited health literacy and low-literate individuals.

Phase 1 Reflections by patients with limited health literacy and low-literate individuals				
Jargon words (frequently used)	**focus group 1 (n=11)** *patients with limited health literacy (reformulation of jargon words)*	**focus group 2 (n=3)** *low-literate individuals (reflection on reformulation)*	**focus group 2** *low-literate individuals (suggestion for extra jargon words to add to the guide)*	
BRCA 1	name of one of the breast cancer genes. The abbreviation is from Breast-Cancer. A mistake in this gene causes an increased risk of breast cancer and ovarian cancer	important to explain the difference between BRCA 1 and BRCA 2 and the connection between the illness and change in a gene	BRCA 1	
BRCA 2	name of one of the breast cancer genes. The abbreviation is from Breast-Cancer. A mistake in this gene causes an increased risk of breast cancer and ovarian cancer (risk of ovarian cancer is lower than with a BRCA 1 mutation)		BRCA 2	
Cells	'building blocks' of your body	unclear that DNA is in the cells	Cells	
Clinical geneticist	physician with a specialization in heredity	**	Clinical geneticist	
			To diagnose*	to determine if someone has a disease (e.g. breast cancer)
DNA	this contains all your personal characteristics. It's your blueprint or the recipe of your body	important to explain abbreviations. DNA is associated with police work. Blueprint is a clear description.	DNA	
DNA-test	a test to find out if there are any changes in your DNA	**	DNA test	
Familial breast cancer	when breast cancer is common in the family	**	Familial breast cancer	
Family history	the diseases that are in the family	**	Family history	

Phase 1 Reflections by patients with limited health literacy and low-literate individuals			
Jargon words (frequently used)	**focus group 1 (n=11)** *patients with limited health literacy (reformulation of jargon words)*	**focus group 2 (n=3)** *low-literate individuals (reflection on reformulation)*	**focus group 2** *low-literate individuals (suggestion for extra jargon words to add to the guide)*
Family tree	a drawing of your family and relatives; father, mother, brothers, sisters, grandparents and so on	**	Family tree
Gene	a small piece of your DNA with a special characteristic, like the color of your eyes	**	Gene
Genetic counselor	someone who gives information and advice about heredity and genetic testing	the difference between geneticist and counselor is difficult. Don't use foreign words.	Genetic counselor
Gene mutation	change or mistake in a gene, in a piece of DNA	most difficult word	Gene mutation
Gene panel	a group of genes investigated at the same time	panel is associated with a group of individuals	Gene panel
Genetic predisposition	if a certain disease is in your family and you can pass it on to the next generation	**	Genetic predisposition
Genetic test	hereditary test	addition: DNA-test	Genetic test
Hereditary	something your parents pass on to you; it is 'in the family'	**	Hereditary
Hereditary screening	testing to find out if a certain disease is in your family	**	Hereditary screening
Inheritance	how the disease is passed on within the family	**	Inheritance
Increased risk	you are more likely to get the disease	increased risk is just 'bad luck'	Increased risk
Mamma care	breast care in the hospital	explain the term 'mamma'. It is associated with being pregnant, breastfeeding	Mamma care

Table 2. Continue

Phase 1 Reflections by patients with limited health literacy and low-literate individuals				
Jargon words (frequently used)	**focus group 1 (n=11)** *patients with limited health literacy (reformulation of jargon words)*	**focus group 2 (n=3)** *low-literate individuals (reflection on reformulation)*	**focus group 2** *low-literate individuals (suggestion for extra jargon words to add to the guide)*	
Mamma surgeon	a surgeon who is specialized in breast-care and cancer	**	Mamma surgeon	
Mutation	a change or a mistake	most difficult to understand	Mutation	
Mutation carrier	someone with a change or a mistake in one of the genes	it is helpful to use this word in a sentence.	Mutation carrier	
			Pathologic examination*	examination of tissue and cells in a laboratory
			Risk factor*	something that increases the chance of getting a disease
Screening	medical exam to find out if there is an abnormality	screening is associated with preventive	Screening	
Transmissible	something in the family that can be passed on to the next generation, such as a disease, or eye color	**	Transmissible	
			Tumor*	benign of malignant (cancer) growth

* *missing words added by target group of patients based on the information letter.*
** *clear description.*

Table 3. Background characteristics of healthcare professionals who responded to the questionnaire.

n=66		**n**	**%**
Sex	male	7	10.6%
	female	59	89.4%
Discipline	breast surgeon	5	7.6%
	specialized nurse	24	36.4%
	physician assistant	4	6.0%
	clinical geneticist	15	22.7%
	genetic counselor	7	10.6%
	other	11	16.7%

the reflection and reformulation by patients with limited health literacy and the reflection on the draft of the guide by low-literate individuals.

Outcomes phase 2: Intended end-users' feedback

Of the 106 healthcare professionals invited to participate, 66 responded (62%) of whom 58 completed the entire online questionnaire (55% of those invited). Table 3 shows background characteristics of healthcare professionals who responded to the questionnaire.

Almost 17% of healthcare professionals indicated that certain words on the preliminary list were unnecessary and almost 27% of them said that specific words in relation to breast cancer genetic counseling were missing. Their reflections were based on daily practice during breast cancer genetic counseling. Healthcare professionals also evaluated the plain-language guide on completeness, usefulness and acceptance. They considered six words in the guide to be unnecessary and they suggested that 11 words be added to the guide. According to the healthcare professionals the following words were unnecessary: *familial breast cancer, genetic test, genetic counselor, gene panel, family tree and mamma surgeon*. They suggested that the following words be added: *autosomal dominant inheritance, HER 2 positive, mammography, MRI, physician assistant, preventive examination, specialized nurse, receptor, sentinel lymph node, triple negative tumor, other breast cancer genes (like CHEK2, PALPB2, ATM)*.Based on daily practice and experience during the Erfo4all training sessions, the project team decided how to adapt the guide, in accordance with these suggestions.

More than half (57%) of the healthcare professionals stated that they had the intention to use the plain-language guide predominantly in consultations with patients with limited health literacy or a migrant background. Almost 65% of the healthcare professionals stated they would share the plain-language guide with colleagues. Suggestions for adaption of the guide mostly concerned content and design, for example digitalizing the guide or to providing it in a pocket-sized format. Table 4 shows the perceived usefulness of the plain-language guide.

Table 4. Perceived usefulness of the plain-language guide for genetic counseling and testing (GenGuide) of healthcare professionals who completed the questionnaire.

n=58	GenGuide facilitates the start of a conversation about GCT (%)	GenGuide will benefit patients (%)	Genguide seems effective for discussing GCT (%)	GenGuide seems a useful addition to my work (%)	GenGuide seems time saving (%)	GenGuide seems easy to use (%)	Intention to use the GenGuide frequently (%)
totally agree	13.8	31.0	13.8	13.8	3.4	12.1	6.9
agree	43.1	58.6	44.8	55.2	27.6	62.1	50.0
neutral	8.6	8.6	22.4	13.8	20.7	12.1	19.0
not agree	24.1	1.7	12.1	8.6	36.2	6.9	17.2
totally disagree	10.3	-	6.9	8.6	12.1	6.9	6.9

Plain-language guide for genetic counseling and testing

The result of the input from patients, low-literate individuals and intended end-users, is a plain-language guide for healthcare professionals (clinical geneticists, genetic counselors and breast surgeons) with 33 jargon words reformulated in a clear and concise description in plain language.

DISCUSSION

In this study we developed a plain-language guide based on clinical practices and tailored to the needs and preferences of patients with limited health literacy and low literate individuals. Based on their input and preferences an elaborate list of jargon words was reformulated in plain language. This is useful because when communicating with patients, healthcare professionals have a tendency to use medical jargon. Avoiding the use of medical jargon, and instead using plain language can overcome important barriers in discussing breast cancer genetic counseling and testing.. Such a guide might help healthcare professionals discuss (referral to) breast cancer genetic testing in a more comprehensible way. This is not only important for patients with limited health literacy or low literacy, but in communication with all patients. Especially because most healthcare professionals experience difficulties in recognizing limited health literacy[19].

Other studies have described the development of a plain language support tool for cancer clinical trials or plain language summaries of scientific articles[23,40] and found that this could play an important role in the patient-physician dialogue. However, these studies were merely focused on patient empowerment and not directly on improving communication behavior from healthcare professionals. To our knowledge this is the first plain-language guide in context of genetics, developed with a focus on healthcare professionals' behavior.

Table 5. Final version of the plain-language guide for genetic counseling and testing.

Jargon word	Plain language
BRCA 1	name of one of the breast cancer genes. The abbreviation is from Breast-Cancer. A mistake in this gene causes an increased risk of breast cancer and ovarian cancer
BRCA 2	name of one of the breast cancer genes. The abbreviation is from Breast-Cancer. A mistake in this gene causes an increased risk of breast cancer and ovarian cancer (risk of ovarian cancer is lower than with a BRCA 1 mutation)
Cells	'building blocks' of our body
CHEK 2	name of one of the breast-cancer genes. A mistake in this gene causes an increased risk of breast cancer, but this risk is lower than with the BRCA 1 and BRCA 2 genes.
Clinical geneticist	physician with a specialization in heredity
Diagnose	to determine if someone has a disease (e.g. breast cancer)
DNA	this contains all your personal characteristics. It's your blueprint or the recipe of your body
DNA-test	a test to find out if there are any changes in your DNA
Familial breast cancer	when breast cancer is common in the family
Family history	the diseases that are in the family
Family tree	a drawing of your family and relatives; father, mother, brothers, sisters, grandparents and so on
Gene	a small piece of your DNA with a special characteristic, like the color of your eyes
Genetic counselor	someone who gives information and advice about heredity and genetic testing
Gene mutation	change or mistake in a gene, in a piece of DNA
Gene panel	a group of genes investigated at the same time
Genetic predisposition	if a certain disease is in your family and you can pass it on to the next generation
Genetic test	heredity test, DNA test
Hereditary	something your parents pass on to you; it is 'in the family'
Hereditary screening	testing to find out if a certain disease is in your family
Increased risk	you are more likely to get the disease
Inheritance	how the disease is passed on within the family
Mamma care	breast-care in the hospital
Mammography	X-ray of the breasts
Mutation	a change or a mistake
Mutation carrier	someone with a change or a mistake in one of the genes
Pathologic examination	examination of tissue and cells in a laboratory
Physician assistant	healthcare professional who independently takes over medical tasks from the clinical geneticist
Preventive examination	a medical examination to see if there are indications of a disease, such as breast cancer
Risk factor	something that increases the chance of getting a disease
Screening	medical exam to find out if there is an abnormality
Transmissible	Something in the family that can be passed on to the next generation such as a disease or your eye color
Triple negative tumor	a special type of breast cancer, the tumor has special characteristics
Tumor	benign or malignant (cancer) growths

Moreover, in the previous studies reformulation in plain-language was not based on preferences and suggestions from patients with limited health literacy or low health literate individuals.

Study limitations

Methodological considerations of our study mainly concern the selection of jargon words for the preliminary list. This selection was based on suggestions of healthcare professionals and not generated by listening to actual encounters with patients with limited health literacy. This may be a shortcoming of our study,however, the frequently used jargon words on the list were derived from the Erfo4all group training sessions together with breast surgeons and specialized nurses. Based on eight training sessions, these jargon words were considered to be representative. In the process of rephrasing jargon words, the focus was on the input from patients with limited health literacy and low-literate individuals. Healthcare professionals just reflected on the draft of the guide for practical implications and to increase the chance of a successful implementation. The intention of healthcare professionals to use the guide was relatively low (57%). We didn't ask healthcare professionals to explain their answer in an open ended question, so a valid explanation for the low intention to use rate is unclear, which is a shortcoming of our study. However, the perceived usefulness of the guide was high, so we are confident that more healthcare professionals will actually use the guide after implementation in daily practice.

The group of healthcare professionals that completed the questionnaire consisted mostly of clinical geneticists and specialized nurses. As breast surgeons were underrepresented in this study, the results on the usefulness and acceptability of the guide may not be entirely representative for this group. However, the feedback from specialized nurses who closely work together with the surgeons can be considered as a reflection of the acceptability of the plain-language guide in routine cancer care.

Practice implications

The use of plain language can improve communication with patients with limited health literacy and provides opportunities for these patients to make informed decisions to participate in genetic testing. Our plain-language guide could improve communication about genetic testing with patients with limited health literacy among a diverse group of healthcare professionals involved in breast cancer care. Surgeons and specialized nurses discuss referral to genetic counseling with eligible breast cancer patients and after referral clinical geneticists and genetic counselors discuss genetic testing and the possible consequences. As genetic testing becomes further integrated into oncology, surgeons and medical oncologists are increasingly discussing the options and possible outcomes of genetic testing with patients, and request these tests themselves. This results in a growing need among healthcare professionals involved in breast cancer care to

communicate genetics information and facilitate decision making in a short time frame[41]. Discussing the consequences of genetic testing with patients with limited health literacy is time-consuming. Our plain-language guide is expected to be helpful to discuss genetic counseling and testing with these groups of patients more effectively.

We believe that the process for development of a plain-language guide can be translated to other health care context, because most of the terminology used in healthcare can be confusing for patients, especially for patients with limited health literacy or at times of distress when people may struggle more than usual to take in information[42,43]. For implementation in daily practice we will take into account the suggestions from healthcare professionals to digitalize the guide and to provide the guide in a pocket-sized format.

Research recommendations

It seems feasible to develop a plain-language guide based on frequently used jargon words in daily practice and reformulate these words based on preferences and understanding from patients with limited health literacy and low-literate individuals. Future research should focus on testing the plain-language guide in a real-world setting and on the effect on patient activation and making informed decisions about participating in cancer genetic counseling and testing. Although other studies suggest that health literacy affects decision making in healthcare, more research is needed on how the use of plain language and specifically how a plain-language guide for healthcare professionals may influence the decision-making process to participate in (breast) cancer genetic testing. It might be interesting to explore opportunities to make the plain-language guide available for patients.

Next to the use of jargon or technical terminology, also other language characteristics of the medical dialogue, such as general language complexity or dialogue pacing, density and interactivity play a role in patients' understanding about genetic information[25]. It is worthwhile to take these into consideration for future research. Finally, although the plain-language guide was well received by intended end-users, we have not yet assessed the actual use in daily practice. It would be interesting to find out if assessment of patients' literacy level with the REALM-G[28] prior to medical consultation will contribute to the use of the plain-language guide.

CONCLUSION

In this study we described the development process of a plain-language guide for breast cancer genetic counseling. Our study showed that reformulation of frequently used jargon words in breast cancer genetic counseling and testing, together with patients with limited health literacy and low-literate individuals, is feasible. The result is a plain-language guide for healthcare professionals to discuss breast cancer genetic counseling in words that are

understandable for these groups of patients. The collaboration with breast cancer patients in the reformulating process, provides valuable insights into plain language synonyms from patients' perspective. Furthermore, lay views often differ from those of patients and healthcare professionals, so reflection on the plain-language guide by low-literate individuals with lay knowledge provided an extra check on the formulation and comprehensibility of the guide.

Reluctance on the part of healthcare professionals to use a new tool is a risk in implementation. In the development of the plain-language guide intended end-users (specialized nurses, breast surgeons, clinical geneticists and genetic counselors,) were actively involved. They brought in frequently used words, evaluated the guide, reflected on a draft version and rated the guide regarding its usefulness and acceptability. The plain-language guide appears to be acceptable and useful, so implementation in daily practice in genetics as well as in mainstream oncology services seems worthwhile and feasible. This is important, because patients are increasingly urged to become involved in decision making, like the decision to participate in genetic counseling and testing. Therefore, attention for health literacy deficits, by using plain language, by speaking in words easily understood by patients, is a necessary first step.

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Authors' contributions

MA and JG conceived and designed the study. ER performed the focus group interviews. SvD and JG analyzed the data. MA and JG developed the plain-language guide. All authors reflected on the questionnaire. JG wrote the paper and all authors revised it critically and approved the final submitted version.

Compliance with ethical standards

All procedures followed were in accordance with the ethical standards of the responsible clinic. For this study, no ethical approval was required. Participants of the (focus) group interviews gave permission to audio-record the interview for study purposes.

Conflict of interest

Jeanine van der Giessen, Margreet Ausems, Els van Riel, Adam de Jong, Mirjam P. Fransen and Sandra van Dulmen declare that they have no conflict of interest.

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PART III

Evaluation of a health literacy training program and its effect on disparities in referral to breast cancer genetic counseling and testing

CHAPTER 5

5

Communication about breast cancer genetic counseling with patients with limited health literacy or a migrant background: evaluation of a training program for healthcare professionals

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Abstract

Access to breast cancer genetic counseling is suboptimal for patients with limited health literacy or a migrant background due to ineffective communication and lack of healthcare professionals' recommendation. This study examines the effect of a blended training program (Erfo4all) for healthcare professionals on their awareness, knowledge and self-efficacy towards communication about genetic counseling with patients with limited health literacy or a migrant background.

In total 59 breast surgeons and specialized nurses from 16 Dutch hospitals completed an online module and group training. Knowledge, self-assessed awareness and self-efficacy were assessed before the training and 33 participants also completed a posttest questionnaire six months after the training program. We also assessed the perceived applicability and relevance of the training program from healthcare professionals' perspectives.We found a significant increase in self-assessed awareness of the prevalence and impact of limited health literacy and in healthcare professionals' self-efficacy to recognize limited health literacy and to communicate effectively with patients with limited health literacy or a migrant background. We didn't find an increase in knowledge score. Almost all healthcare professionals reported that they use the techniques learned in the training, such as the teach-back method andplain language, and felt more confident discussing breast cancer genetic counseling. Our results suggest that a blended training program for healthcare professionals has potential to improve their ability to communicate effectively about breast cancer genetic counseling with patients with limited health literacy or a migrant background and offers a promising way to increase the referral rate for these groups of patients.

Key words: breast cancer, referral, genetic testing, health literacy, blended training program.

INTRODUCTION

Breast cancer, the most commonly diagnosed type of cancer, affects about 12% of women worldwide[1]. Between 5- and 10% of breast cancers are associated with a hereditary predisposition. Pathogenic variantsin the BRCA1 and BRCA2 genes are associated with about 20% of familial breast and ovarian cancers[2]. Female breast cancer patients with a BRCA1 or BRCA2 pathogenic varianthave an increased risk of a second primary breast cancer and/or ovarian cancer. A pathogenic variantmay also affect healthy family members, so genetic testing of breast cancer patients at risk of carrying a pathogenic variantis important[3-5]. Healthcare professionals (surgical oncologists, medical oncologists and specialized nurses) should identify patients at risk of carrying a variantin a breast cancer gene (e.g. BRCA1, BRCA2, CHEK2, PALB2, ATM genes), inform them about genetic testing, refer them for genetic counseling or request a test themselves. Yet, despite the fact that genetic testing has been available for over two decades, many eligible women do not receive BRCA testing[6] and for certain groups of patients the substantial underutilization of genetic testing is even larger[7]. Previous research has shown that there is unequal access to cancer genetic testing for patients with lower educational levels and those with a migrant background[8-12]. The major cause for this seems to lie in the absence of surgeon recommendations[13,9]. Referral is not always adequately discussed with these groups of patients and limited health literacy as well as cultural differences seems to play a role[8,12,14,15]. This might be related to ineffective communication which is widely recognized as a major contributor to health disparities[15]. Health literacy refers to the skills to get access to, understand, appraise and use health-related information in various domains[16]. Estimations of the prevalence of limited health literacy in the Netherlands range between 29% - 36% in the general population.[17,18], 24.4 % of Dutch population has a migrant background, from which 10.5% has a western migrant background and 13.9% has an non-western background[19]. For effective referral, it is important that surgeons and specialized nurses, the main referrers to genetic counseling for patients with breast cancer[20,8], possess adequate awareness, knowledge and skills to identify patients with limited health literacy[21].

Healthcare professionals seem to be insufficiently aware of the negative impact of limited health literacy on medical communication, failing to recognize limited health literacy in patients and lacking the skills to effectively discuss referral to breast cancer genetic counseling[22,23]. They might benefit from a training in recognizing these patients and in discussing referral to genetic counselling adequately in order to optimize access to genetic care for these groups of patients. As limited health literacy, cultural factors and language proficiency are interrelated, training programs should attend to literacy problems as well as to cultural differences[24,25].

Training program for healthcare professionals

We developed a blended training program (Erfo4all) based on healthcare professionals' and patients' needs and preferences, to increase healthcare professionals' knowledge, awareness and self-efficacy when communicating about breast cancer genetic counseling with communication-vulnerable patients, i.e. patients with limited health literacy or a migrant background[26].

In our blended training program, we selected an integrated approach to health literacy and communication with patients with a migrant background. The training program was developed within the Erfo4all project, a project that aims to achieve equal access to breast cancer genetic counseling for all eligible patients. The program consisted of two successive parts: an online knowledge-module (20 minutes) and a group training in one of the participating hospitals (2 hours). The online module included information about the prevalence of low literacy and limited health literacy in the Netherlands, the relevance of health literacy in understanding and appraising information from healthcare professionals as well as the way health literacy relates to socio-economic and background characteristics of patients. It also included information about how to communicate effectively with patients with a migrant background. In the group training, the teach-back method - a methodology used by healthcare providers to check whether a patient understands what has been discussed - was used as a technique to identify patients with limited health literacy and role-play was used as a strategy to acquire required communication skills and to practice the use of plain language[27-29]. To further enhance healthcare professionals' cultural competences, cultural sensitivity training techniques were introduced. The training was led by a professional trainer from Pharos (Dutch Centre of Expertise on Health Disparities) and role play sessions, based on real life cases, were practiced with a training actress.

The aims of the current study were to assess:

1. The extent to which this training program increased:
   a. healthcare professionals' knowledge and awareness about the impact and prevalence of limited health literacy, awareness of the impact of limited health literacy on medical communication, their self-efficacy to recognize patients with limited health literacy and to communicate effectively about breast cancer genetic counseling.
   b. b) healthcare professionals` awareness of the importance of taking into account cultural factors when communicating with patients with a migrant background and their self-efficacy in coping with thesecultural factors and with language barriers.
2. The perceived applicability and relevance of the training program from healthcare professionals' perspective.

METHODS

A total of 73 healthcare professionals involved in breast cancer treatment from 19 hospitals in the Netherlands responded to an invitation to participate in the Erfo4all training program. Before the start of the training program, they were asked to fill out an initial online questionnaire (T0) to get access to the online knowledge module. Within two weeks they were invited to attend a group training on location. We used a pre-/posttest design to evaluate the effect of the Erfo4all intervention on knowledge, self-assessed awareness and self-efficacy related to communication about breast cancer genetic counseling with patients with limited health literacy or a migrant background.Six months after the group training the healthcare professionals were asked to fill out a second questionnaire (T1), again to assess knowledge, awareness and self-efficacy. At T1 we added self-report questions concerning the applicability and relevance of the training program as well as healthcare professionals' perceived awareness regarding the problem of health literacy in general and their confidence to adapt their communication effectively. Only healthcare professionals who completed the whole program and filled out the T0 and T1 questionnaire, were considered in the pre- and post-intervention analysis. For the additional questions, all healthcare professionals who filled in the T1 questionnaire were included.

Outcome variables

Outcome variables were assessed with self-constructed online questionnaires, before (T0) and six months after the training (T1). The items in these questionnaires were based on the intervention mapping approach and the matrix of change used for the development of our training program.[26]

Knowledge was assessed with five multiple choice questions focusing on: the prevalence of low literate adults in the Netherlands, limited (health) literacy among people with a migrant background, the prevalence of adults with limited health literacy in the Netherlands, the level of education in relation to the level of health literacy, and option of using of a professional interpreter (self- reported). Each item was rated as correct (1) or incorrect (0) and a total knowledge score was computed as the number of correct answers.

Awareness was assessed by three items: prevalence and impact of health literacy in the Netherlands, impact of health literacy on medical communication, and the importance of taking into account cultural factors when communicating with patients with a migrant background.

Items were scored on a five-point Likert scale ranging from (1) hardly aware to (5) very highly aware.

Self-efficacy was assessed by five statements on having confidence in: recognizing limited health literacy in patients, communicating effectively about breast cancer genetic counseling with patients with limited health literacy, understanding which customs and

habits from patients with a migrant background might influence communication, coping with cultural factors in communication with patients with a migrant background and coping with a language barrier.

Items were scored on a 5-point Likert scale ranging from (1) totally disagree to (5) totally agree.

At T1 we also assessed healthcare professionals' perceived awareness regarding the problem of limited health literacy in general and their confidence to tailor communication about breast cancer genetic counseling to the needs of communication-vulnerable patients. These additional items were scored on a 5-point Likert scale ranging from (1) totally disagree to (5) totally agree. The relevance of the training was assessed by asking whether the trained healthcare professionals shared information about the training with colleagues in multidisciplinary oncology meetings. Application of skills, such as the use of the teach-back method and plain language, was assessed with a five point Likert scale ranging from (1) never to (5) very regularly.

Statistical analysis

Univariate analysis was used to describe the background characteristics of healthcare professionals.

Categorical data, number of healthcare professionals, discipline, sex and clinical setting are presented in numbers and percentages. Continuous data, like age and work experience, are presented as means and standard deviations. To analyze T0 and T1 data, we performed paired analysis or repeated measurement, since multiple responses from the same subject cannot be regarded as independent from each other. In this analysis, subjects were included only when data were available from both time points (T0 and T1). To check for potential selective drop-out we compared the group that only completed the T0 (T0 only) with the group who completed both questionnaires (T0+T1) and looked for differences in demographics and outcome variables. To analyze pre-post differences in the outcome measures knowledge, awareness and self-efficacy we used the Wilcoxon signed-rank for related samples. Tests for statistical significance were two-sided with $\alpha=0.05$. Statistical analyses were performed using SPSS version 24.0 (SPSS Inc, Chicago, IL).

RESULTS

The baseline questionnaire (T0) was completed by 65 participants of whom 59 were working in one of 16 hospitals and completed the whole training program (online module and group training). In total 37 participants filled out the T1 questionnaire, of which 33 participants from 14 hospitals filled out both questionnaires (T0 and T1). Table 1 shows the background characteristics of healthcare professionals who only completed the first questionnaire

Table 1. Background characteristics of healthcare professionals

variable		T0 n=59*			T0-T1 n= 33**		
total		**n**	**%**	**sd**	**n**	**%**	**sd**
sex	male	11	18.6%		7	21.2%	
	female	48	81.4%		26	78.%	
discipline	breast surgeon	17	28.8%		9	27.3%	
	specialized nurse	36	61.0%		20	60.6%	
	medical oncologist	1	1.7%		1	3.0%	
	physician assistant	2	3.4%		2	6.0%	
	other	3	5.1%		1	3.0%	
age			45.8	8.5		45.2	8.7
work experience in breast cancer care (years)			10.7	7.1		11.0	6.0
clinical setting	university hospital	6	10.2%		4	12.1%	
	community hospital	53	89.8%		29	87.8%	

** n=59: group who completed T0 questionnaire and whole training program*
***n=33: group who completed T0 questionnaire, whole training program and T1 questionnaire*

(n=59) and of those who completed both questionnaires (n=33).Based on background characteristics, no statistical differences were found between both groups, indicating that drop out between T0 and T1 was not selective.

Pre-/posttest changes in awareness, knowledge and self-efficacy

Table 2 shows the pre-/posttest scores of the participating healthcare professionals on self-assessed awareness, knowledge and self-efficacy (n=33). At the posttest (after the training), there was a significant positive change on six outcome measures compared to the pretest (before the training). The largest increase was observed in participants' self-efficacy. Understanding the customs and habits of patients with a migrant background, the ability to recognize limited health literacy, to communicate effectively about breast cancer genetic counseling, and to cope with cultural factors or a language barrier, significantly increased from baseline to T1. Moreover, awareness of the prevalence and impact of limited health literacy in the Netherlands increased significantly. The total knowledge score did not increase over time.

Perceived applicability and relevance of the training program

In our prior study we assessed acceptability and usefulness of the training program directly after the group training[26]. Six months after completing the training program we assessed applicability and relevance of the training program in daily practice (n=37). More than 80% (n=30) of the healthcare professionals reported having used plain language to explain genetic testing; of these, almost 41% (n=12) reported using it (very) regularly. Even more (92%, n=34) reported applying the teach-back method to discover whether a patient understood information and to identify limited health literacy, while 62% (n=23) reported

Table 2. Awareness, knowledge and self-efficacy score of healthcare professionals before and after the training

	n	%	n	%	* -	=	+	p-value
Awareness								
awareness of prevalence and impact limited health literacy in the Netherlands					1	17	15	.003*
• low	-	-	-	-				
• barely	1	3.0%	-	-				
• reasonable	28	84.8%	18	54.5%				
• high	3	9.1%	13	39.4%				
• very high	1	3.0%	2	6.1%				
awareness of the impact of limited health literacy on medical communication					5	13	15	.052
• low	-	-	-	-				
• barely	3	9.1%	2	6.1%				
• reasonable	18	54.5%	10	30.3%				
• high	10	30.3%	19	57.6%				
• very high	2	6.1%	2	6.1%				
awareness of the importance to take into account cultural factors					5	21	7	.644
• low	-	-	-	-				
• barely	1	3.0%	-	-				
• reasonable	5	15.2%	5	15.2%				
• high	23	69.7%	24	72.7%				
• very high	4	12.1%	4	12.1%				
Self-efficacy								

	n	%	n	%	* -	=	+	p-value
having confidence to understand which customs and habits from patients with a migrant background might influence communication					2	9	22	.000*
• totally disagree	1	3.0%	-	-				
• disagree	14	42.4%	1	3.0%				
• not agree/not disagree	14	42.4%	16	48.5%				
• agree	3	9.1%	14	42.4%				
• totally agree	1	3.0%	2	6.1%				
having confidence to recognize limited health literacy in patients					4	14	15	.008*
• totally disagree	-	-	-	-				
• disagree	7	21.2%	1	3.0%				
• not agree/not disagree	16	48.5%	14	42.4%				
• agree	10	30.3%	18	54.5%				
• totally agree	-	-	-	-				
having confidence to communicate effectively about breast cancer genetic counselingwith patients with limited health literacy					0	8	25	.000*
• totally disagree	-	-	-	-				
• disagree	13	39.4%	-	-				
• not agree/not disagree	14	42.4%	8	24.2%				
• agree	6	18.2%	24	72.7%				
• totally agree	-	-	1	3.0%				

Table 2. Continued

	n	%	n	%	* -	=	+	p-value
having confidence to cope with cultural factors in communication with patients with a migrant background					0	15	18	.000*
• totally disagree	-	30.3%	-	-				
• disagree	10	39.4%	1	3.0%				
• not agree/not disagree	13	30.3%	5	15.2%				
• agree	10	-	27	81.8%				
• totally agree	-	-	-	-				
having confidence to cope with a language barrier					2	19	12	.024*
• totally disagree	-	-	-	-				
• disagree	5	15.2%	2	6.1%				
• not agree/not disagree	12	36.4%	8	24.2%				
• agree	16	48.5%	21	63.6%				
• totally agree	-	-	1	3.0%				
• missing	-	-	1	3.0%				
Knowledge								
Sum scores / total knowledge Correct answers:					10	11	12	.465
• 1	1	3.0%	-	-				
• 2	5	15.2%	3	9.1%				
• 3	11	33.3%	15	45.5%				
• 4	14	42.4%	11	33.3%				
• 5	2	6.1%	4	12.1%				

using the teach-back method (very) regularly. More than 97% (n=36) of the respondents felt more aware of health literacy problems and assessed their ability to recognize patients with limited health literacy higher and 86% (n=32) reported that their ability to communicate effectively with these groups of patients had improved. Most healthcare professionals expected to benefit from a booster training session after one year and more than 41% (n=15) reported sharing their experience with the training program in multidisciplinary oncology meetings with colleagues.

DISCUSSION

The results of our study suggest that a blended training program for healthcare professionals (i.e. breast surgeons and specialized nurses) aimed at increasing awareness, knowledge and self-efficacy regarding limited health literacy and communication with patients with limited health literacy or a migrant background, leads to significant improvement in awareness of the prevalence and impact of limited health literacy, andself-efficacy in communicating about breast cancer genetic counseling with these groups of patients. No significant differences were found in pre- and posttestknowledge scores, on awareness of the impact of health literacy on medical communication and the importance of taking into account cultural factors when communicating with patients with a migrant background. Almost half (41%) of the breast surgeons and specialized nurses who participated in the training reported to share s their experience with colleagues and almost all reported to apply the techniques taught in the training in daily practice (i.e. teach-back method and using plain language).

Healthcare professionals experience several problems in discussing genetic counseling and testing with patients with limited health literacy or a migrant background. Low awareness of the problem of limited health literacy, difficulties in recognizing limited health literacy, coping with cultural factors and a language barrier in communication with patients with a migrant background and a lack of the skills needed to discuss referral effectively with these patients are the main problems.. This is often compounded by patients' limited health literacy which has been shown to negatively affect their ability to play an active role in their own health care, by asking questions, participating in shared decision making and taking initiative[30]. Health literacy is thus not an individual issue, and making health care accessible by adapting communication to patients' understanding and abilities, is critical[31].

The increase in self-assessed self-efficacy six months after the training is promising, because this outcome variable has found to be associated with actual communication performance[32,33]. Self-efficacy beliefs determine whether certain behavioral change will be initiated and also influences the effort one puts forth to change a certain behavior. For breast surgeons and specialized nurses, an increase in self efficacy when communicating

with communication-vulnerable patients, is a prerequisite to actually changing their communication style. The use of role-play in the group training might have contributed to this result. Based on Bandura's theory on self-efficacy[34], role-play has been described as one of the most effective methods to improve self-efficacy[33].

Communication skills training has previously been shown to produce a significant and durable increase in the self-efficacy of healthcare professionals[35-38]. However, in these interventions training durations varied from a 3.5 hour workshop to a 10-day course, and they were based on traditional face-to-face learning. For future research it would be interesting to discover whether short or long term differences exist between these different approaches.

We were surprised that we didn't find an increase in knowledge score mostly because other studies on health literacy training interventions with a pre-/post survey, showed significant improvements in (perceived) health literacy knowledge[38,39]. Furthermore, a systematic review from Liu showed that blended learning, which combines traditional face-to-face learning and e-learning,has a large consistent positive effect on knowledge acquisition[40]. We therefore expected our blended learning approach to contribute to an increase in knowledge. This discrepancy could possibly be ascribed to the fact that healthcare professionals participated in our training program on a voluntary basis. It is likely that their interest in the subject as well as their motivation to participate in a health literacy training program provide an explanation for the relatively high knowledge scores at baseline. Their motivation to participate also reflects the fact that we, in contrast to other studies, did not highlight a lack of awareness of health literacy at baseline and also that awareness of taking into account cultural awareness was already high, so there may be a ceiling effect[23].

This is one of the few studies examining the effect of a training program for breast surgeons and specialized nurses with a focus on health literacy and also, to our knowledge, the first study explicitly focusing on discussing breast cancer genetic counseling with patients with limited health literacy or a migrant background. Given the high rates of low or limited health literacy in the Netherlands (36.4% of adults)[17], and even more in the rest of Europe[41], health literacy sensitive training interventions could help healthcare professionals communicate with these vulnerable groups of patients.

The dual challenges of limited health literacy and cultural differences are likely to increase due to an expanding and increasingly diverse population[25], so the effect of an integrated approach, with a focus on limited health literacy and cultural differences in one training program, is interesting for future research.

The results however, should also be examined in light of the study's limitations. First, the response rate at T1 was relatively low and this may represent a selection of healthcare professionals who are more inclined to respond. Practical reasons for this low response rate may be related to the large time period that has passed since the training and a lack of tim of the participating healthcare professionals. However, despite this low response rate,

the 33 healthcare professionals who completed both questionnaires do not appear to systematically differ from the group who only filled out the first questionnaire. Therefore, the results can be considered representative, and thus we could extrapolate the results to the whole group and the time interval between pretest and posttest was long enough to avoid a testing effect. Second, as there were no standardized questionnaires available we used a self-constructed questionnaire, developed on the basis of a theoretical approach. Because it has not been validated, it may be subject to measurement error, and conclusions cannot be made with total confidence. Third, as it was impossible to randomly assign participants to groups, we choose a pretest-posttest design without a control group. This lack of a control group made it difficult to control for confounding variables. Finally, the use of self-reported outcome measures on awareness, self-efficacy and applied skills indicate attitudes rather than behavior. There is a risk of social desirability bias because we did not actually observe skills in daily practice, but instead asked healthcare professionals if they (felt able to) apply certain communication skills. Although it is unknown whether the increased scores on awareness and self-efficacy in this case indeed led to sustainable changes in communication behavior in daily practice, feelings of self-efficacy have been linked previously with behavioral change[42]. Future studies should also examine provider-patient communication in the consulting room as we previously did in breast cancer genetic counseling[43].

In conclusion, our study shows improvements in relevant outcome measures among a diverse group of healthcare professionals involved in surgical breast cancer care in different regions in the Netherlands. It is promising that the skills learned during the training seem applicable in daily practice, even in the long term, and that healthcare professionals reported gains in awareness and self-efficacy. We implemented the Erfo4all training program in three regions in the Netherlands in different clinical settings (academic and non-academic hospitals and among healthcare professionals from different disciplines.Our previous study showed that the acceptance and perceived usability of the program was high[26]. Thus, widespread implementation of the training program seems feasible, making it a promising intervention for other healthcare professionals in cancer care. As genetics and genomics become part of mainstream medicine, effective communication about genetic testing becomes even more important with the potential to either reduce or exacerbate disparities in access to genetic testing[44].

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Compliance with ethical standards

All procedures followed were in accordance with the ethical standards of the responsible clinic. The Medical Ethical Committee considered the Dutch Medical Research involving Human Subjects Act not applicable to this study. Therefore, a formal review by the medical ethics committee was not required.

Conflict of interest

The authors declare that they have no conflict of interest.

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CHAPTER 6

6

Effect of a health literacy training program for surgical oncologists and specialized nurses on disparities in referral to breast cancer genetic testing

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Abstract

Background: There is an underuse of genetic testing in breast cancer patients with a lower level of education, limited health literacy or a migrant background. We aimed to study the effect of a health literacy training program for surgical oncologists and specialized nurses on disparities in referral to genetic testing.
Methods: We conducted a multicenter study in a quasi-experimental pre-post (intervention) design. The intervention consisted of an online module and a group training for surgical oncologists and specialized nurses in three regions in the Netherlands. Six months pre- and 12 months post intervention, clinical geneticists completed a checklist with socio-demographic characteristics including the level of health literacy of each referred patient. We conducted univariate and logistic regression analysis to evaluate the effect of the training program on disparities in referral to genetic testing.
Results: In total, 3179 checklists were completed, of which 1695 were from hospital referrals. No significant differences were found in educational level, level of health literacy and migrant background of patients referred for genetic testing by healthcare professionals working in trained hospitals before (n=795) and after (n=409) the intervention. The mean age of patients referred by healthcare professionals from trained hospitals was significantly lower after the intervention (52.0 vs. 49.8, P=0.003).
Conclusion: The results of our study suggest that the health literacy training program did not decrease disparities in referral to genetic testing. Future research in a more controlled design is needed to better understand how socio-demographic factors influence referral to breast cancer genetic testing and what other factors might contribute.

Keywords: Breast cancer genetic testing, referral, access to care, health literacy, training program, communication.

INTRODUCTION

Despite guidelines that recommend genetic testing for patients at increased risk of carrying a pathogenic variant in a breast cancer gene (e.g. *BRCA1*, *BRCA2*, *CHEK2*, *PALB2*, *ATM*) [1-3] not all eligible patients are referred for genetic testing. Previous studies show that patients with a lower level of education or a (non-Western) migrant background have poorer access to genetic testing.[4-9] These disparities in referral may lead to differences in treatment and survival rates, because early detection of a pathogenic variant has the potential to improve health outcomes.[10] Besides, carrying a *BRCA* pathogenic variant implies a change in follow-up measures as these patients may have an increased risk of developing a second breast cancer or ovarian cancer. The detection of a pathogenic variant enables predictive DNA testing in healthy family members. [11-13] Currently, eligible newly diagnosed breast cancer patients are usually offered rapid genetic testing before their primary surgery.[14,15] These patients mostly have a higher overall genetic testing uptake compared to patients in routine care.[16]

Several barriers to genetic testing have been identified, including worries regarding insurance coverage for genetic testing and concerns about misuse of testing, privacy and confidentiality issues.[17,18] In addition, patients with a lower level of education or a migrant background have limited access to genetic testing due to a lack of physician recommendation.[5,7,9,19] Ineffective communication is widely recognized as a major contributor to such health disparities.[20] Patients' level of health literacy, i.e. the degree to which someone has the capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions, seems to play an important role. [21-26] Individuals with limited health literacy may understand less of the written and oral communication they receive about genetic information and may participate less in consultations with healthcare professionals.[25,27] They also have less medical knowledge, which might hamper patient-initiated inquiry.[28] Non-cognitive aspects of health literacy, such as motivation and self-confidence, also defined as 'the capacity to act', are also likely to have an impact on communication, making it difficult for patients to participate actively in healthcare decisions.[29,30] Among patients with a lower level of education or a migrant background, the level of health literacy is relatively low.[31] Besides, limited language proficiency in turn affects the level of health literacy, reduces access to healthcare systems and leads to poorer health outcomes.[32,33]

Surgical oncologists and specialized nurses, the main referrers to genetic testing for patients with breast cancer, may be insufficiently aware of the negative impact of limited health literacy on medical communication.[5,34] They do not recognize limited health literacy in patients and lack the skills needed to effectively discuss (referral to) breast cancer genetic testing.[24,35] We therefore developed a health literacy training program (Erfo4all) for healthcare professionals (i.e. surgical oncologists and specialized nurses involved in breast cancer care),

consisting of an online module and a group training on location.[36] In a previous study, the effect of a health literacy program on healthcare professionals' awareness, knowledge and self-efficacy related to communication about genetic testing with patients with limited health literacy or a migrant background was examined.[37] The program appeared to improve healthcare professionals' ability to communicate effectively about breast cancer genetic testing with 'communication-vulnerable' patients.[38,39] The overall aim of the current study was to evaluate the effect of the health literacy training program on disparities in referral to breast cancer genetic testing. Specific research questions were:

1. What are the background characteristics of all patients referred by healthcare professionals in trained hospitals compared to those of patients referred in untrained hospitals?
2. a) Does the number of patients with a lower level of education, limited health literacy or a migrant background referred by healthcare professionals from trained hospitals differ before and after the health literacy training program? b) Do these numbers vary between the rapid genetic testing setting and routine care?

METHODS

Study design

We used a quasi-experimental pre-post (intervention) design to study the effect of the health literacy training program. Healthcare professionals from 19 hospitals (4 academic and 15 non-academic hospitals), who refer patients for breast cancer genetic testing to one of the four university medical centers in three regions in the Netherlands, were invited to participate in a health literacy training program.[36,37] The training program consisted of an online module (18 min) and a group training on location (2 h.). The online module focused on knowledge acquisition, while in the group training practicing skills were most important.[36]

Participants

To measure the effect of the health literacy training program on the rates of referral, clinical geneticists and genetic counselors from the four university medical centers were asked to fill in a checklist for all new patients referred for breast cancer genetic testing. They started with the checklist registration approximately 6 months before the training of healthcare professionals (baseline) in their region and continued until 12 months after the training. The total registration period in the study was from March 2017 until March 2019 Inclusion in the pre- or post-intervention group was based on (estimated) date of referral. All breast cancer patients who were treated in academic and non-academic hospitals, and referred for diagnostic genetic testing by their surgical oncologist or specialized nurse, were eligible

for the study. Patients referred by their general practitioner were excluded because general practitioners were not invited to the training program. These patients were mainly referred for predictive genetic testing (e.g. testing when a pathogenic variant was detected in an affected family member).

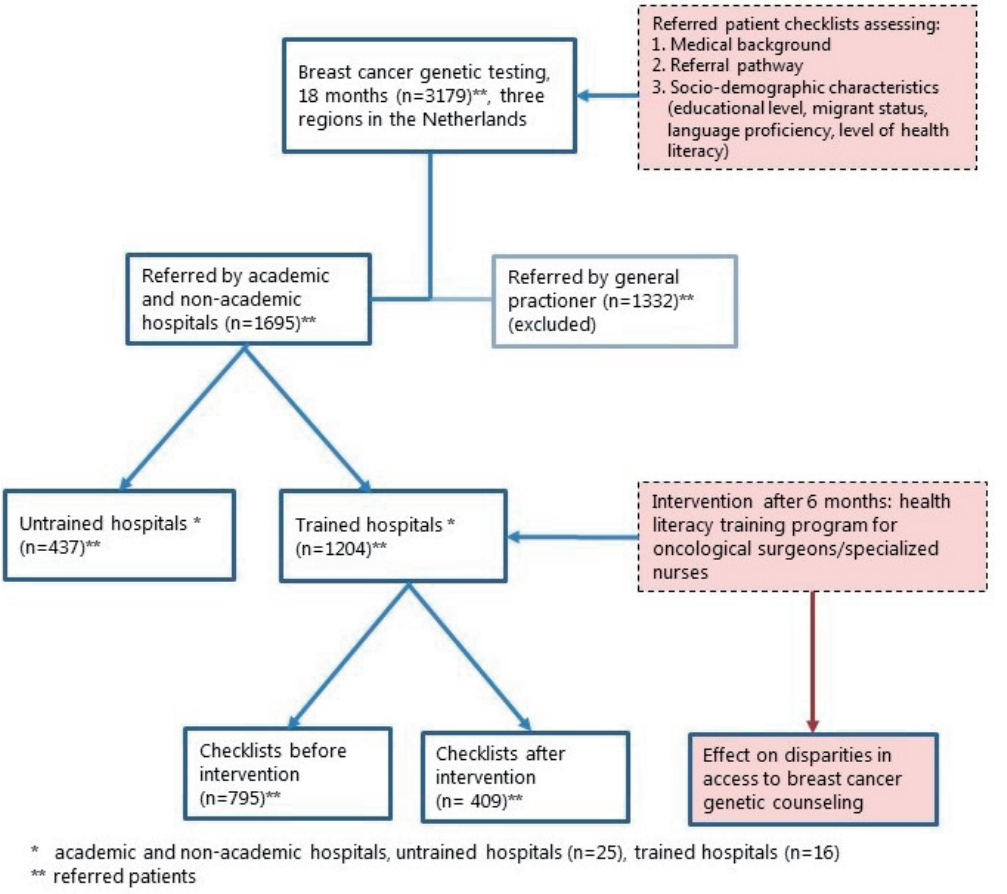

Figure 1. Study design health literacy training program.

Data collection

Checklist

The checklist used in this study was based on previous studies on determinants of referral to breast cancer genetic testing.[5,6] The checklist contained patients' demographics (i.e. level of education, migrant status, level of health literacy, language proficiency, disease status and referral pathway (i.e. referred by general practitioner or a hospital), referral for diagnostic or predictive DNA testing, and referral for rapid genetic testing or routine care).

Patients' level of education was determined by the Dutch Standard Classification of Education [40] and the international classification of the UNESCO [41], i.e. lower level of education: (pre-) primary education or first stage of basic education; intermediate-1 educational level: lower secondary or second stage of basic education; intermediate-2 educational level: (upper) secondary education; and higher level of education: tertiary education. The migrant status of the counselee was determined according to the definition of Statistics Netherlands.[42] According to this definition, a patient is a migrant when at least one of their parents was born outside of the Netherlands. Furthermore, a distinction was made between Western migrants (at least one parent born outside the Netherlands, but in Europe, North America, Australia, New Zealand, Indonesia or Japan) and non-Western migrants (at least one parent was born in Turkey or countries in Africa, Latin America or Asia). Because of practical considerations (time constraints) and ethical considerations, it was not possible to ask patients to complete one of the health literacy assessment instruments during the visit at the outpatient clinic, like the Short Test of Functional Health Literacy in Adults (S-TOFHLA) or the Rapid Estimate of Adult Literacy in Medicine (REALM). To still get an indication of the level of health literacy, we choose a valid measurement that was most likely to be applicable in everyday clinical practice. The level of health literacy was assessed by one of the validated screening questions from Chew known to be effective in identifying patients with inadequate health literacy, i.e.: 'How often do you have someone help you read hospital materials?'[43]

Trained hospitals

A total of 73 healthcare professionals from 19 hospitals that were invited, responded to the invitation. Healthcare professionals (n=59) from 16 hospitals completed the whole training program. However, not all healthcare professionals working in one of the 16 hospitals and referring patients to breast cancer genetic testing, participated in the training program. We assumed that the trained healthcare professionals shared their learning experience during multidisciplinary meetings and therefore use the term 'trained hospitals' to indicate healthcare professionals from hospitals that participated in the training program. We previously showed that more than 41% of the healthcare professionals actually reported to share their experience with their colleagues.[37] Healthcare professionals referring from 'control' hospitals are defined as 'untrained hospitals'(n=25). Only patients referred by healthcare professionals in a trained hospital were considered in the analyses for the pre- and post-intervention comparison. In the analysis, we furthermore distinguished between rapid referrals, i.e., early after diagnosis when results are needed for treatment plans, and routine referrals. Due to privacy issues, it was not always possible to know the actual referral date for patients in routine care. When the actual referral date was unknown, the average waiting time during the registration period was imputed to estimate the referral date. Figure 1 shows the study design.

Statistical analysis

The primary outcome was the percentage of patients referred for breast cancer genetic testing with a lower level of education, limited health literacy or a migrant background. Categorical variables were described as totals and percentages. Continuous variables were described as a mean and standard deviation (SD). Univariate analysis was performed to compare the distribution of patient characteristics before and after the intervention in the trained hospitals, using the independent sample t-test for continuous variables, and Chi-square or Fisher's exact tests for the categorical variables. Patient characteristics included age, breast cancer patient status, migrant status, level of education, and language proficiency. Furthermore, to adjust for potential confounders, such as age, migrant status, referral for rapid counseling and educational level, we performed a multivariate logistic regression. We tested whether it was more likely for women with limited health literacy to be part of the post-intervention group as compared to the pre-intervention group (outcome measure). As language proficiency and limited health literacy were strongly correlated, language proficiency was excluded from the logistic regression model to avoid multi-collinearity. Limited health literacy, with as variable 'need help reading hospital materials', was coded as never/once in a while (0) and often/always (1). All tests were two-sided and $p < 0.05$ was considered statistically significant. The analyses were conducted with SPSS version 24.0 (IBM Corporation, Armonk, NY, USA).

Results

Background characteristics of patients referred for genetic testing

Between March 2017 and March 2019, clinical geneticists and genetic counselors completed 3179 checklists. About half of the referred patients (52%) were affected with breast cancer and 53% of all patients were offered predictive DNA testing. Most referrals (56%) came from hospitals, 44% of the patients were referred by their general practitioner and 45% of the hospital referrals concerned rapid genetic testing. Background characteristics of all patients referred by hospitals (n=1695) showed that the majority of patients had a Dutch background (79%), while 10% of patients had a non-Western migrant background. In total 37% of the patients seen for genetic testing had a high level of education, while 4% had a low level of education. Almost 4% of patients referred by hospitals had low or limited health literacy, and the level of language proficiency was low for 3% of the patients. There were 1204 patients (71%) referred by healthcare professionals from trained hospitals, and 437 patients (26%) by untrained hospitals. We found no differences in background characteristics of patients between the three regions (Utrecht, Amsterdam, Rotterdam). Table 1 shows the background characteristics of all patients referred for genetic testing and those referred by trained and untrained hospital.

Table 1. Background characteristics of all patients referred for genetic testing and patients referred by trained and untrained hospitals

	All counselees		**Hospital referrals***					
			total		**trained hospitals**		**untrained hospitals**	
	N	%	N	%	**N**	**%**	N	**%**
total**	3179		1695					
total excluding missing trained status	--		1641		1204	71.0%	437	25.8%
mean age referral	48.9		51.6		51.2		52.8	
(min-max)	(18 - 92)		(18 - 88)		(18 - 88)		(23 - 83)	
gender	3179		1695		1204		437	
male	294	9.2%	36	2.1%	29	2.4%	7	1.6%
female	2885	90.8%	1659	97.9%	1175	97.6%	430	98.4%
breast cancer	3179		1641		1204		437	
affected	1641	51.6%	1417	86.3%	1024	85.0%	393	89.9%
unaffected	1538	48.4%	224	13.7%	180	15.0%	44	10.1%
mean age breast cancer	49.3		49.2		49.1		49.6	
(min-max)	(24 – 87)		(24 - 87)		(24 - 87)		(27 - 83)	
DNA-testing	3010		1535		1132		403	
diagnostic	1421	47.2%	1178	76.6%	833	73.6%	345	85.6%
predictive	1589	52.8%	357	23.3%	299	26.4%	58	14.4%
rapid DNA-testing	3140		1614		1191		423	
yes	662	21.1%	627	44.6%	475	39.9%	152	35.9%
no	2478	78.9%	987	55.4%	716	60.1%	271	64.1%
educational level	3021		1668		1148		417	
low	103	3.4%	61	3.9%	45	3.9%	16	3.8%
intermediate-I	616	20.4%	353	22.6%	233	20.3%	120	28.8%
intermediate-II	1097	36.3%	566	36.2%	404	35.2%	162	38.8%
high	1205	39.9%	585	37.4%	466	40.6%	119	28.5%

	All counselees		**Hospital referrals***					
			total		**trained hospitals**		**untrained hospitals**	
	N	%	N	%	**N**	**%**	N	**%**
need help because of limited HL	3133		1614		1184		430	
never	2825	90.2%	1448	89.7%	1052	88.9%	396	92.1%
once in a while	199	6.4%	105	6.5%	87	7.3%	18	4.2%
often	47	1.5%	26	1.6%	19	1.6%	7	1.6%
always	62	2.0%	35	2.2%	26	2.2%	9	2.1%
language proficiency	3148		1624		1193		431	
good/intermediate proficiency	3057	97.1%	1574	96.9%	1151	96.5%	423	98.0%
low proficiency	52	1.6%	28	1.7%	24	2.0%	4	1.0%
no proficiency	39	1.2%	22	1.4%	18	1.5%	4	1.0%
migrant status counselee	3146		1623		1192		431	
Dutch native	2550	81.1%	1289	79.4%	927	77.7%	362	84.0%
migrant	596	18.9%	334	20.6%	265	22.2%	69	16.0%
country of origin known	575		324		259		65	
☐ western	307	9.8%	164	10.0%	134	11.2%	30	6.7%
☐ non western	268	8.5%	160	9.6%	125	10.5%	35	8.1%
interpretor present	3150		1625		1193		432	
no	3100	98.4%	1593	98.0%	1169	98.0%	424	98.1%
yes	50	1.6%	32	2.0%	24	2.0%	8	1.9%
☐ family	42	87.5%	29	90.6%	21	87.5%	0	0.0%
☐ professional	6	12.4%	3	9.4%	3	12.5%	8	100.0%

* excluding records training unknown

** hospital referrals n =1695 (53%) and general practitioner referrals n = 1332 (42%), unknown n=152 (5%)

Effect of the health literacy training program on disparities in referral to breast cancer genetic testing in routine care and rapid genetic testing

For 729 patients in the Utrecht region the date of referral could be retrieved. For 966 patients from the other two regions, we could only register the week or month of first consultation at the genetics department. Among the 1204 breast cancer patients referred by healthcare professionals in trained hospitals, 795 (66%) breast cancer patients were referred before the intervention and 409 (44%) after the intervention. In the univariate analysis for the pre- and post-intervention groups, no significant association was found between migrant status, level of education, or level of health literacy and the intervention.

Looking at health literacy, we found that 89 (11.4 %) breast cancer patients with low or limited health literacy are referred before the intervention and 43 (10.7%) were referred after the intervention. Moreover, multivariate regression analysis showed no effect on referral to genetic testing of patients with limited health literacy after the introduction of the health literacy training program (OR=0.399, 95% CI=0.156-1.021), after adjusting for potential confounding factors such as age, migrant status, referral for rapid counseling and level of education. Moreover, no difference was found in the separate analyses between rapid genetic testing only (OR=0.69, 95% CI=0.25-1.92) and routine care only (OR=0.69, 95% CI=0.27-1.74). In addition, lower age was statistically significantly associated with the intervention, indicating that younger patients were more likely to be referred for genetic testing after the intervention (p=0.003). This effect was not found in patients who underwent rapid genetic counseling. Table 2 shows pre- and post-intervention results for all patients referred by healthcare professionals from trained hospitals and the results of patients referred for rapid genetic testing and in routine care.

Unexpected results

Due to the significant increase in the self-efficacy of the trained healthcare professionals to communicate effectively with patients with limited health literacy or a migrant background found previously,[37] we were surprised that our current study showed no effect on the referral rate of these groups of patients. As sample bias might have been introduced in the pre-intervention group, we conducted an additional logistic regression analysis with untrained hospitals as a second pre-intervention group. With data from this additional analysis, the referral rate of migrant patients tended to be higher (p=0.063) in trained hospitals after the intervention as compared to referral rate in untrained hospitals. Table 3 shows the result of the logistic regression analyses with patients referred by trained hospitals as the pre-intervention group and the additional logistic regression analysis with patients referred by untrained hospitals as the pre-intervention group.

Table 2. Pre- and post- intervention results of breast cancer patients referred by healthcare professionals from trained hospitals in routine care and rapid genetic counseling

Variable		Total		Before intervention		After intervention		p-value
		N	%	N	%	N	%	
ALL REFERRALS TRAINED HOSPITALS								
Total		1204		795		409		
Mean age referral (min – max)		51.2 (18 – 88)		52.0 (18 – 88)		49.8 (20 – 87)		**0.003***
affected (N=1204)	yes	1024	85%	680	85.5%	344	84.1%	**n.s.**
	no	180	15%	115	14.5%	65	15.9%	
migrant status counselee (N=1192)	Dutch native	927	77.8%	610	77.8%	317	77.5%	**n.s.**
	Migrant	265	22.2%	173	22.2%	92	22.5%	
	• Western	134	50.6%	80	46.2%	54	58.7%	**n.s.**
	• Non western	125	47.2%	89	51.4%	36	39.1%	
	• unknown	6	2.3%	4	2.3%	2	2.2 %	
educational level (N=1148)	low	45	3.9%	33	4.4%	12	3.0%	**n.s.**
	intermediate-I	233	20.3%	163	21.6%	70	17.7%	
	intermediate-II	404	35.2%	264	35.1%	140	35.4%	
	high	466	40.6%	293	38.9%	173	43.8%	
need help because of limited HL (N=1184)	never	1052	88.9%	694	88.6%	358	89.3%	**n.s.**
	once in a while	87	7.3%	57	7.3%	30	7.5%	
	often	19	1.6%	11	1.4%	8	2.0%	
	always	26	2.2%	21	2.7%	5	1.2%	
language proficiency (N=1193)	good/fair	1151	96.5%	756	96.2%	395	97.1%	**n.s.**
	bad	24	2.0%	17	2.2%	7	1.7%	
	none	18	1.5%	13	1.7%	5	1.2%	
interpretor present (N=1193)	no	1169	98,0%	768	98.0%	401	98.0%	**n.s.**
	yes	24	2.0%	16	2.0%	8	2.0%	
family or professional known (N = 24)	• family	21	87.5%	14	87.5%	7	87.5%	
	• professional	3	12.5%	2	12.5%	1	12.5%	

Table 2. Continued

Variable		Total		Before intervention		After intervention		p-value
		N	%	N	%	N	%	
RAPID GENETIC COUNSELING ONLY								
Total		475		302		173		
Mean age referral (min – max)		46.3 (18 – 76)		46.3 (18 – 75)		46.4 (25 – 76)		**n.s.**
migrant status counselee (N=469)	Dutch native	355	75.7%	226	76.4%	129	76.4%	**n.s.**
	Migrant	114	24.3%	70	23.6%	44	25.4%	
	• Western	40	8.5%	18	3.8%	22	4.7%	
	• Non western	70	14.9%	49	10.4%	21	4.4%	
educational level (N=454)	low	15	3.3%	11	3.9%	4	2.4%	**n.s.**
	intermediate-I	81	17.8%	50	17.5%	31	18.3%	
	intermediate-II	153	33.7%	91	31.9%	62	36.7%	
	high	205	45.2%	133	46.7%	72	42.6%	
need help because of limited HL (N=465)	never	408	87.8%	261	88.5%	147	865%	**n.s.**
	once in a while	38	8.2%	21	7.1%	17	10.0%	
	often	7	1.5%	4	1.4%	3	1.8%	
	always	12	2.6%	9	3.1%	3	1.8%	
language proficiency (N=469)	good/fair	455	97.0%	287	96.6%	168	97.7%	**n.s.**
	bad	6	1.3%	4	1.3%	2	1.2%	
	none	8	1.7%	6	2.0%	2	1.2%	
interpretor present (N=470)	no	461	98.1%	291	98.0%	170	98.3%	**n.s.**
	yes	9	1.9%	6	2.0%	3	1.7%	
family or professional known (N=9)	• family	8	88.9%	5	83.3%	3	100.0%	
	• professional	1	11.1%	1	16.7%	0	0.0%	

Variable		Total		Before intervention		After intervention		p-value
		N	%	N	%	N	%	
NON-RAPID REFERRALS ONLY								
Total		716		486		230		
Mean age referral (min – max)		54.6 (20 – 88)		55.6 (22 – 88)		52.5 (20 – 87)		**0.002***
migrant status counselee (N=710)	Dutch native	561	79.0%	378	78.8%	183	79.6%	**n.s.**
	Migrant	149	21.0%	102	21.3%	47	20.4%	
	Western	94	13.2%	62	8.7%	32	4.5%	
	Non western	53	7.3%	39	5.5%	14	2.0%	
educational level (N=682)	low	30	4.4%	22	4.8%	8	3.6%	**n.s.**
	intermediate-I	149	21,8%	110	23.8%	39	17.7%	
	intermediate-II	245	35.9%	170	36.8%	75	34.1%	
	high	258	37.8%	160	34.6%	98	34.6%	
need help because of limited HL (N=706)	never	632	89.5%	427	88.8%	205	91.1%	**n.s.**
	once in a while	48	6.8%	35	7.3%	13	5.8%	
	often	12	1.7%	7	1.5%	5	2.2%	
	always	14	2.0%	12	2.5%	2	0.9%	
language proficiency (N=711)	good/fair	683	96.1%	462	95.9%	221	96.5%	**n.s.**
	bad	18	2.5%	13	2.7%	5	2.2%	
	none	10	1.4%	7	1.5%	3	1.3%	
interpretor present (N=710)	no	695	97.9%	470	97.9%	225	97.8%	**n.s**
	yes	15	2.1%	10	2.1%	5	2.2%	
family or professional known (N=15)	• family	13	86.7%	9	90.0%	4	80.0%	
	• professional	2	13.3%	1	10.0%	1	20.0%	

Table 3. Logistic regression analysis with differences between trained and untrained hospitals as pre-intervention group

	Pre-and post- intervention trained hospitals			**Untrained hospitals as pre-intervention group**		
Variable	**odds ratio**	**95% CI**	**p-value**	**odds ratio**	**95% CI**	**p-value**
health literacy	0.399	0.156 -1.021	0.055	0.707	0.217 -2.307	0.565
migrant status	1.113	0.816 -1.517	0.500	1.428	0.981 -2.080	0.063
rapid genetic counseling	0.906	0.696 -1.179	0.461	0.883	0.653 -1.194	0.419
mean age referral	0.988	0.977 -0.999	0.026*	0.984	0.984 -0.996	0.010*

CONCLUSIONS

Our study did not find an effect of a health literacy training program for surgical oncologists and specialized nurses on disparities in referral of patients with a lower level of education, limited health literacy or a migrant background. There were no differences in referral in the rapid genetic counseling setting. In general, the uptake in this setting is already higher compared to routine care because a DNA test may influence surgical treatment decisions. Healthcare professionals believe that rapid genetic testing is beneficial for patients and therefore the tendency to refer eligible patients might be higher.[11]

An important finding of our study was that the health literacy training program could make a difference for younger patients with breast cancer in routine care. Referral for the group of younger patients is important because young age at diagnosis of breast cancer indicates a higher risk to carry a *BRCA1* or *BRCA2* pathogenic variant and is a clear indication for referral to breast cancer genetic testing.[44] Despite this, physicians do not systematically discuss genetic testing with young women with breast cancer.[8,45] Therefore, there was extra attention in the training program for the importance of the referral of young (migrant) patients with breast cancer.

Our study has some clear strengths. We conducted a multicenter study, and the involvement of different genetic departments in three regions in the Netherlands increased the generalizability of our study. Further, we included almost 3200 checklists with medical and socio-demographic information of breast cancer patients, of which 1695 checklists (from hospital referrals) are included in analysis. This large sample size is large enough to draw conclusions.

Next to the strengths, there are limitations. It is important to reconsider the study design of the health literacy training program. The most important limitation of our study is the fact that it is unknown which patients are **not** referred during the registration period. We could only register the percentage of referred patients with a lower level of education, limited health literacy or a migrant background. This makes a difference in interpreting the

results. Second, based on practical and ethical implications, it was not possible to register for each counselee the healthcare professional who referred the patient to the department of genetics. Instead we used the hospital (trained or untrained) as an independent variable. Third, due to the relatively small number of patients with limited health literacy and a migrant background, there might be a sample fluctuation of patients referred by trained hospitals that are included in the study. The additional logistic regression analysis confirmed that the pre-intervention group might not be representative, which may (partly) explain the unexpected results of our study. Next, the exact date of referral was unknown for patients referred in two regions, so we could not conclude with 100% certainty that referral took place before the intervention. To correct for this omission, we imputed the referral date based on average waiting time. Finally, we used the validated question of Chew (i.e., 'How often do you have someone help you read hospital materials?) as a self-reported measure to determine the level of health literacy. Although Chew showed that this single question may identify individuals with inadequate health literacy,[43] respondents may have given socially desirable answers or may have been too embarrassed to admit that help is needed with reading or interpreting medical information.

Implications for future research

Future research, using a more controlled design, with a larger sample size of patients with limited health literacy or a migrant background is needed to further investigate disparities in referral to breast cancer genetic testing. Furthermore, valid measurement of patient's level of health literacy is important. For healthcare professionals, being able to correctly assess the patient's level of health literacy is a prerequisite for effective communication.

Next to factors on the side of the healthcare professionals, like competences to communicate effectively with patients with limited health literacy, other factors might contribute to referral to genetic counseling. Patient's request, for example, also impacts the referral to breast cancer genetic counseling. Yet, taking the initiative for referral is difficult for patients with limited health literacy. They more often consent to providers' recommendation.[46]

Despite the fact that our study showed no effect on referral to breast cancer genetic testing, we believe in the importance of effective communication and improving the communication skills of healthcare professionals. For all interventions designed to reduce disparities in access to genetic testing and testing, communication about genetic testing in a comprehensible way, for instance by using plain language and using the teach-back method, is an important condition.[47,48] Especially when genetic testing becomes part of mainstream medicine – with the potential to make genetic services accessible to all eligible patients – adapting communication about genetic testing to patients' needs and abilities is even more essential.[49]

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Conflict of interest statement
The authors declare no conflict of interest.

Compliance with ethical standards.
No ethical approval was required. The Medical Ethical Committee of the University Medical Center Utrecht considered the Dutch Medical Research involving Human Subjects Act, not applicable to this study.

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CHAPTER 7

Summary and general discussion

In this final chapter, the main results of this thesis are summarized, followed by a discussion of these findings for each part of the thesis. Subsequently, a reflection on the methodological considerations and implications for clinical practice and future research is given.

Patients with a lower educational or migration background seem to have poorer access to breast cancer genetic counseling and testing. Referral is not always adequately discussed with these groups of patients, and limited health literacy as well as cultural differences seem to play a role.[1-4] Ineffective communication is recognized as one of the contributing factors to differences in access to healthcare and to health disparities in general.[5] Health disparities have been defined as differences in health that are closely linked with social, economic, and/or environmental disadvantage.[6] Health literacy is commonly associated with many of the antecedents of health disparities, including access to care.[7,8] The exact relationship between limited health literacy and access to care in general remains unclear, but in the context of genetics, provider recommendation seems important.[2,9-11]

The general aim of the research project described in this thesis was to develop, implement and evaluate an intervention to reduce disparities in referral to breast cancer genetic counseling and testing. We assessed educational level and migrant status of counselees referred to cancer genetic counseling (part I) and developed a health literacy training program and a tool to communicate effectively with communication-vulnerable patients (part II). In part III the effect of the health literacy training program on awareness, knowledge and self-efficacy of healthcare professionals and on the referral rate of patients with limited health literacy or a migrant background is described.

SUMMARY OF MAIN FINDINGS FOR EACH PART

Part I - Disparities in referral to (breast) cancer genetic counseling and testing

To determine educational level and migrant status of counselees referred to cancer genetic counseling, we assessed personal characteristics and demographics of 731 counselees, including 209 breast cancer patients. The results of this study are described in chapter 2. In comparison with the Dutch population, less counselees with a lower and intermediate-2 educational level and more highly educated counselees participated in cancer genetic counseling and testing. In addition, there were less migrants compared to the general population ($p<0.001$). In a previous study at the department of genetics of the University Medical Center Utrecht, an underrepresentation in cancer genetic counseling of migrant patients and patients with a lower educational background was found.[2]

The results of the current study, seven years after the first study, show that this underrepresentation has not changed since then. The research in part I of this thesis provided evidence that there is still unequal access to (breast) cancer genetic counseling and testing for patients with a lower educational background and migrant patients,

indicated as communication-vulnerable patients. These disparities in referral may lead to differences in treatment and survival rates, because early detection of a pathogenic variant in a cancer gene has the potential to improve health outcomes.[12] Other studies confirm these disparities in access to (breast) cancer genetic counseling and testing.[2,9,11,13-15] Several barriers to genetic counseling and testing have been identified, including worries regarding insurance coverage for genetic testing and concerns about misuse of testing results, privacy and confidentiality issues.[16,17] However, next to these factors, physician recommendation seems most important. Various studies show that lack of physician recommendation is the primary reason many eligible breast cancer patients remain untested.[2,9,15,18] Most patients who do not participate in genetic testing, have never discussed genetic testing with their healthcare professional.[19]

Part II - Development of a health literacy training program and a plain-language guide.

The aim of the studies in the second part of this thesis was to develop a health literacy training program for healthcare professionals and a tool to communicate effectively with patients with limited health literacy or a migrant background. The systematic development of the health literacy program based on patients' and healthcare professionals' needs and preferences is described in chapter 3. Prior to the training we assessed awareness, knowledge and self-efficacy of healthcare professionals. Directly after the training, acceptability and usefulness of the program were assessed. The outcome of this study was a health literacy training program (Erfo4all) consisting of an online module (18 min.) and a group training on location (2 hrs.). Although healthcare professionals showed moderate to high awareness about prevalence and impact of limited health literacy and cultural factors, they did not feel confident in recognizing limited health literacy. Also, their self-efficacy to communicate effectively with these communication-vulnerable patients was low. The training program was evaluated positively by the healthcare professionals. They reported a high degree of acceptance, and the combination of an online module and a group training on location was considered useful and time-efficient.

The development of a plain-language guide to communicate effectively with patients with limited health literacy or a migrant background is described in chapter 4. In this study, we drew up a list of jargon words that are frequently used verbally and in writing during breast cancer genetic counseling consultations. Input was given by surgical oncologists and specialized nurses who participated in the Erfo4all training program, and a clinical geneticist and a genetic counselor from the department of genetics of the University Medical Center Utrecht. Subsequently, we conducted a focus group interview with breast cancer patients with a lower educational background or limited health literacy and a personal experience with breast cancer genetic counseling, and reformulated these words in plain language. The draft of this guide was evaluated in a second focus group interview with low-literate

individuals with no personal experience in breast cancer genetic counseling. We also aimed to explore intended end-users' (surgical oncologists, specialized nurses, clinical geneticists, genetic counselors) perceptions of the plain-language guide on completeness, acceptability and usefulness. Our study showed that reformulation of jargon words in breast cancer genetic counseling and testing is feasible. The input from patients with limited health literacy and low-literate individuals provided valuable insights in plain-language synonyms. The result was a plain-language guide for healthcare professionals (clinical geneticists, genetic counselors and breast surgeons) with 33 jargon words reformulated in a clear and concise description in plain language. Acceptability and perceived usefulness of the guide among these intended end-users was high.

The results described in chapter 3 are supported by other studies on communication with patients with limited health literacy or a migrant background. Healthcare professionals have difficulties in recognizing patients with limited health literacy and their self-efficacy in adapting their communication accordingly, is low.[20-22] This indicates a need for techniques to communicate effectively with patients with limited health literacy or a migrant background. This need for training, specifically on communication with patients with limited health literacy, has been reported by others as well.[20,21,23] The results of chapter 3 show that it is feasible to develop a health literacy training program in the context of cancer genetic counseling based on training needs and preferences of healthcare professionals and tailored to patients' perspectives. The value of using plain-language in communication with patients is clear.[24, 25] However, when communicating with patients, healthcare professionals tend to use medical jargon.[26] Avoiding medical jargon and using plain language instead, can overcome important barriers in discussing breast cancer genetic counseling and testing. Other studies have described the development of a plain-language support tool for cancer clinical trials or plain-language summaries of scientific articles and found that this could play an important role in the patient-physician dialogue.[27,28] However, these studies were merely focused on patient empowerment in a research setting and not directly on improving communication behavior of healthcare professionals and shared decision making. A plain-language guide might help healthcare professionals to discuss (referral to) breast cancer genetic testing in a more comprehensible way. This is not only important for patients with limited health literacy or low literacy, but in communication with all patients. Assuming that all patients may have difficulty comprehending medical information and getting access to healthcare, calls for a universal health literacy precautions approach.[29,30]

Part III - Evaluation of a health literacy training program and its effect on disparities in referral to breast cancer genetic counseling and testing

The effect of a health literacy training program for healthcare professionals on their awareness, knowledge and self-efficacy towards communication about genetic counseling and testing with patients with limited health literacy or a migrant background is described

in chapter 5 of this thesis. In total 59 surgical oncologists and specialized nurses from 16 Dutch hospitals completed an online module and group training. Knowledge, awareness and self-efficacy were assessed before the training and 33 participants also completed a post-test questionnaire six months after the health literacy training program. No significant differences were found in knowledge scores and on awareness of the impact of health literacy on medical communication and the importance to take into account cultural factors when communicating with patients with a migrant background. However, we found a significant increase in self-assessed awareness of the prevalence and impact of limited health literacy and in healthcare professionals' self-efficacy to recognize limited health literacy and to communicate effectively with patients with limited health literacy or a migrant background. Although training durations vary, which makes it difficult to compare, other studies also show that communication skills training programs produce a significant and durable increase in the self-efficacy of healthcare professionals.[31-33] As mentioned in chapter 3, directly after the group training, healthcare professionals rated the training program as acceptable and useful. Six months after completing the training program, healthcare professionals who participated in the program evaluated the health literacy training program positively. They reported a high degree of acceptance with the blended learning method; the combination of an online module and a group training on location was considered useful and time-efficient. Almost all healthcare professionals who filled out the post-questionnaire (n=33) reported that they use the techniques learned in the training, such as the teach-back method and plain language, and felt more confident discussing breast cancer genetic counseling with patients with limited health literacy or a migrant background. More than 41% of the healthcare professionals that participated in the training program reported sharing their experience with the training program in multidisciplinary oncology meetings with colleagues.

Chapter 6 describes the effect of the Erfo4all training program on referral of patients with limited health literacy or a migrant background to breast cancer genetic counseling and testing. We used a quasi-experimental pre-post (intervention) design to study the impact of the health literacy training program on referral of these communication-vulnerable patients. No significant differences were found in educational level, level of health literacy and migrant background of breast cancer patients referred by healthcare professionals working in trained hospitals. Before the intervention (n=795) breast cancer patients were referred and after the intervention (n=409) breast cancer patients . The mean age of patients referred by healthcare professionals from trained hospitals was significantly lower after the intervention (52.0 vs. 49.8, P=0.003). With data from additional logistic regression analysis with untrained hospitals as a second pre-intervention group, the referral rate of migrant patients tended to be higher. No difference was found in referral of patients with limited health literacy.

There has been increased research interest in the field of health literacy and the impact of health literacy on the relationship between patient and healthcare professional.[21,34,35] The health literacy training program developed within the Erfo4all program, seems to be effective on healthcare professionals' awareness of the problem of limited health literacy and their self-efficacy in communication with patients with limited health literacy or a migrant background. The significant increase in self-efficacy of healthcare professionals, as described in chapter 5 of this thesis is consistent with the findings of others.[32,33,36] These studies also showed a durable increase in self-efficacy of healthcare professionals as a result of a (mandatory) health literacy training program. In line with the conclusion in our study, it remains unclear whether the increased self-efficacy led to observable changes in communication behavior. The increase in health literacy knowledge as described by others, was not consistent with the findings in our study.[37,38] This discrepancy could possibly be ascribed to the fact that healthcare professionals participated in our training program on a voluntary basis, they showed interest in the subject and their knowledge score at baseline was already relatively high. Though health literacy training for healthcare professionals seems important, it is still an underdeveloped domain in the health professions education field.[39,40] However, improving skills of healthcare professionals to effectively communicate with patients with limited health literacy in order to help them to take deliberate decisions to improve their health, is necessary. Therefore, creating opportunities for health literacy training as part of continued medical education (CME), as well as integrating health literacy training in the education of medical students, is important. Other studies show that interventions with an integrated approach, combining knowledge with skill acquisition, developed in real-world settings together with patients and healthcare professionals, tend to be most successful.[39] Also, health literacy training programs with a multidimensional approach, like including cultural competences or organizational demands, are considered to be important.[30,39,41,42] The Erfo4all training program meets these conditions and therefore we expect successful implementation to be possible.

Unexpectedly, the Erfo4all training program did not reduce disparities in access to breast cancer genetic counseling and testing for limited health literate patients and migrant patients. In our study, and based on our conceptual model on referral to breast cancer genetic counseling, we choose to focus on medical consultations and specifically on healthcare professionals' communication skills. For that reason, we developed a health literacy training program for healthcare professionals. We are aware that other factors might play a role in explaining unequal access to breast cancer genetic counseling and testing. Other studies investigating barriers and facilitators to (*BRCA*) genetic counseling in minority populations, described patients' awareness, cost-related factors, stress and distrust and family concerns as contributing factors.[43]. Also, several studies confirm that patients' initiative is important in the referral process and asking questions about genetic testing increases the likelihood of being referred for genetic counseling.[44-47] Lower educated patients and migrant patients

show less initiative and express other information needs for genetic care than higher educated patients. This argues for a more personalized approach in communication about (referral to) breast cancer genetic counseling.[48] Finally, organizational factors are important in relation to access to care. The degree to which hospitals implement strategies to make it easier for patients to understand health information, find their way around the hospital, participate in the healthcare process, and manage their health, might also contribute to access to care for communication-vulnerable patients.

Methodological considerations

The following considerations refer to the adequacy of the methods used in the studies described in this thesis. The overall strengths and limitations on study design, development of the health literacy training program and the plain-language guide are discussed, as well as considerations on the definition and measurement of health literacy.

Study design

We used a quasi-experimental pre-post (intervention) design to study the effect of the Erfo4all training program on referral to breast cancer genetic counseling and testing. To measure the effect of the training program (intervention) on the rates of referral, clinical geneticists and genetic counselors from four university medical centers were asked to fill in a checklist for all new patients referred for breast cancer genetic testing (six months before the intervention and 12 months after). An important strength of our study is that we were able to include medical and socio-demographic information of a large number of patients (n= 3179) of which 1695 were from hospital referrals. Although we were able to compare referrals in three different regions, there are limitations of our study design. The most important limitation is the absence of a control group. Not having a control group cannot eliminate the possible impact of other variables that might have influenced referral. Another important limitation is the fact that it is unknown which patients are **not** referred during the registration period. Nor did we have data about patients who declined referral for breast cancer genetic counseling. Looking at the healthcare professionals, it was unclear if a healthcare professional who referred a patient to the department of genetics participated in the Erfo4all training program. It was not permitted to register the name of the healthcare professional referring a patient, so we could only register if the referring healthcare professional worked in a hospital that had participated in the Erfo4all training program. Therefore, we used the term trained hospitals to indicate healthcare professionals who participated in the training program. We thereby assumed that trained healthcare professionals shared their learning experience in multi-disciplinary oncology sessions with colleagues. This assumption was based on the fact that, in our study on the evaluation of the Erfo4all training program, more than 41% of the healthcare professionals that participated in the training, reported sharing their experience with the training program in

multidisciplinary oncology meetings with colleagues. In addition, the study team organized a kick-off meeting in each participating hospital, often as part of a regular multidisciplinary oncology meeting of the breast cancer team. Non-trained healthcare professionals could have participated in this meeting and therefore have become aware of the referral disparities. It is difficult to estimate the effect on referral behavior of healthcare professionals who did not participate in the training program, but are aware of the problem of underreferral. This can mean both an overestimation and an underestimation of the effect of the health literacy training program. After all, we do not know exactly if and how the information was shared with untrained healthcare professionals and how many healthcare professionals this involves. Finally, due to practical and organizational reasons, the exact date of referral to the genetics department by the surgical oncologist was not always known. This made it difficult to conclude if referral actually took place before the intervention and might have influenced our results. When the actual referral date was unknown the average waiting time during the registration period was imputed to estimate the referral date.

Development of the health literacy training program and the plain-language guide
A strength in the development of the health literacy training program was our systematic approach. We used the intervention mapping method in order to make deliberate decisions regarding the format and content of the program. The group interviews with healthcare professionals ((surgical) oncologists, specialized nurses) and breast cancer patients were found to be a good method to identify their needs and preferences. We experienced that involving healthcare professionals and patients is a promising strategy in the development of a training program. A potential limitation might be the selection of patients for the group interview. Although we aimed for seven patients, we were able to include only three patients. This was due to last minute cancelations by four of the invited patients.

In the development of the plain-language guide the collaboration with breast cancer patients with limited health literacy in the reformulating process was an important strength. This provided valuable insights into plain-language synonyms from patients' perspective. Furthermore, reflection of low literate individuals with lay views provided an extra check on the formulation and comprehensibility of the plain-language guide. Methodological considerations of this part of the study mainly concern the selection of jargon words for the preliminary list. This selection was based on suggestions of healthcare professionals and not generated by listening to actual consultations with patients with limited health literacy or by inquiring about their experiences. Furthermore, surgical oncologists were underrepresented in the group that completed the questionnaire, so the results on the usefulness and acceptability of the guide may not be entirely representative for them.

Definition and measurement of health literacy
In our study we used the validated question of Chew (i.e. 'How often do you have someone

help you read hospital materials?) as a self-reported measure to determine the level of health literacy. This decision was based on practical (time constraints) and ethical considerations for measuring health literacy in clinical practice. The question of Chew gives an indication of the level of health literacy, but respondents may have given socially desirable answers or may have been too embarrassed to admit that help is needed with reading or interpreting medical information. However, defining health literacy is still a point of discussion which also limits the possibilities for valid measurement and comparison.[49] For example, the consideration to not only include cognitive factors and skills, but also psychological factors, like motivation.[50] In addition, Nutbeam proposed a model of health literacy that distinguishes three levels of health literacy: 1) functional health literacy, i.e., basic reading and writing skills to be able to understand and use health information, 2) interactive health literacy, referring to more advanced cognitive and literacy skills to interact with healthcare providers and the ability to interpret and apply information to changing circumstances, and 3) critical health literacy, which refers to more advanced cognitive skills to critically analyze information to exert greater control over one's life.[51,52] Moreover, context-specific literacy is considered relevant. There is increasing recognition of the importance of individuals' knowledge and skills to being able to understand and use genetic and genomic information, referred to as 'genomic literacy'.[53-55] In our study, we used the question of Chew to determine whether a patient was health literate. Therefore it was not possible to differentiate between low, limited or high health literacy nor in the different levels of health literacy (functional, communicative or interactive). This might play a role in interpreting the results.

Implications for clinical practice

Healthcare professionals (surgical oncologists and specialized nurses, n=59) from sixteen hospitals participated in the health literacy training program. This about 21% of the hospitals in the Netherlands offering breast cancer care. Healthcare professionals from hospitals in other regions in the Netherlands remain untrained. The research described in this thesis showed that the acceptance and perceived usability of the Erfo4all program was high, and skills learned during the training seem applicable in daily practice, even in the long term. Moreover, it seems feasible to adapt our health literacy training program as well as the plain-language guide to the context of other hereditary cancers, like gynecological cancer or colon cancer. This makes widespread implementation worthwhile. As mainstream genetic testing, i.e. the implementation of genetic testing in routine care, becomes part of daily practice, communication about genetic testing by (surgical) oncologists and specialized nurses becomes even more important. In order to better meet the needs of communication-vulnerable patients, the use of the teach-back method and plain language must be seen as a core competency in the education of medical students, but also as part of continued medical education programs for healthcare professionals. We propose a stepwise approach for making the Erfo4all training program accessible in medical education as well as for

healthcare professionals working in daily practice. A first step is making the Erfo4all training program accessible for other surgical oncologists and specialized nurses who did not participate in our study. The Dutch Society of Surgical Oncology and the Dutch Professional Nurse Practitioner Organization can play a role in inviting these healthcare professionals for training. The next step is adapting the Erfo4all program for other hereditary cancers and making this training program accessible for other disciplines. For undergraduate students, health literacy training should be a standard part of the curriculum. The effect of our study on communication with patients with limited health literacy, can be used to make targeted choices regarding the content of this curriculum. Finally, the health literacy training could become part of the online introduction program of the hospital for new healthcare professionals and on digital training platforms.

Assessing patients' level of health literacy

In general, and also confirmed in our study, healthcare professionals seem to be insufficiently aware of the negative impact of limited health literacy on medical communication, fail to recognize limited health literacy in patients and lack the skills to effectively discuss referral to breast cancer genetic counseling.[20,56,57] Assessing the level of health literacy of patients can provide the healthcare professional with information about the patients' average level of health literacy. However, there is no 'one size fits all' and there are several instruments, used for various reasons, in different contexts and with different goals. In fact, each situation may invoke the need for a different type of instrument to measure health literacy.[58]

In addition, in clinical reality, practical and ethical considerations often play a role. It is important to take that into account when drawing conclusions about the level of health literacy of patients. In fact, it might be more appropriate to use plain-language and apply the teach-back method for all patients. This method appears beneficial in recognizing limited health literacy and creates an opportunity for effective communication. The universal health literacy approach makes the discussion if assessment of level of health literacy and a targeted approach is needed, relevant.[59]

Directions for future research

Further research on the effect of health literacy training programs

Research on the effect of health literacy training programs on reducing disparities in access to care, is still an underdeveloped domain. For a better understanding of the role of health literacy training programs on reducing these disparities, research in a more controlled design, with targeted interventions and a clear definition of (the measurement) of health literacy is needed. To find out whether health literacy training programs can change healthcare professionals' observable communication behavior, it is useful to examine patient-provider communication in the consulting room (in everyday practice). This is important to find out if, and how, healthcare professionals apply skills in daily practice and what aspects need

further encouragement or training. In addition, implementation research to find out how successful implementation of health literacy training programs in clinical practice can take place, is important.

Further research on other factors influencing access to breast cancer genetic counseling and testing

There are other factors that influence referral and uptake of breast cancer genetic counseling and testing. Not only physician-related factors play a role, but also patient-related factors, like an active patient role and taking initiative for referral. Health literacy is known as a potential risk factor in the communication about genetic counseling and testing. Patients need skills to understand genetic information, to communicate effectively and express their experiences and preferences. It may be worthwhile to consider whether increasing patients' empowerment (e.g., to learn them to ask questions, or take the initiative to discuss possible genetic causes of their breast cancer) can also contribute to effective communication about breast cancer genetic counseling and testing and to an informed decision to participate. Recent studies show promising results in the development and implementation of decision aids in cancer care, adapted to patients with limited health literacy.[60, 61] The use of these decision aids seems promising and should be further explored. In addition, the discussion about the definition of health literacy is currently led by the conviction that health literacy is not only determined by individuals' skills and abilities, but also by the complexities of the healthcare system.[62] Organizational factors are important, which calls for creating more health literate organizations.[63] It is interesting to find out if the transition to a health literate organization, as introduced by the US institute of Medicine is worthwhile in relation to equal access to care.[64]

GENERAL CONCLUSION

In this thesis, the development and testing of an intervention to reduce disparities in referral to breast cancer genetic counseling for patients with limited health literacy or a migrant background is described. Our research shows that these patients, defined as communication-vulnerable patients, have limited access to breast cancer genetic counseling and testing presumably due to a lack of physician recommendation. Training healthcare professionals (surgical oncologists and specialized nurses) is promising to improve communication about breast cancer genetic counseling and testing. Healthcare professionals experience difficulties in recognizing limited health literacy and adapting their communication to the needs of patients with limited health literacy or a migrant background. The results of our research suggest that a health literacy training program can have a significant effect on awareness and self-efficacy of healthcare professionals in communication with these groups

of patients. Applying teach-back and using plain language seem promising strategies in improving effective communication about breast cancer genetic counseling and testing with communication-vulnerable patients. Despite that, we did not find a decrease in disparities in referral to breast cancer genetic counseling and testing after participating in the Erfo4all training program. Methodological considerations, like study design and defining and measuring health literacy may play a role. Moreover, action is required on multiple levels, not only on the side of the healthcare professional, but also on the side of patients and the organization. Systematic development of health literacy training programs for healthcare professionals is needed, as well as more research on the effect of these training programs on communication in healthcare and specifically on disparities in access to genetic testing. Linking health literacy to diversity and inclusion in healthcare is necessary for providing equal access to genetic testing. This starts with recognizing and acknowledging the problem, followed by a targeted approach.

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APPENDICES

A

Appendices

Nederlandse samenvatting/Dutch summary

Acknowledgments/Dankwoord

About the author

Nederlandse samenvatting

Introductie

Borstkanker is wereldwijd één van de meest gediagnosticeerde vormen van kanker; het treft ongeveer 12% van de vrouwen. In de Europese Unie is borstkanker verantwoordelijk voor 29% van de kanker diagnoses bij vrouwen, met een hogere incidentie in de meer welvarende landen. Bij ongeveer 5-10 % van de vrouwen met borstkanker speelt een erfelijke oorzaak een grote rol. De meeste erfelijke vormen van borstkanker worden veroorzaakt door een mutatie in de borstkankergenen *BRCA1* en *BRCA2*. Dragers van een *BRCA1*- of *BRCA2*-genmutatie hebben een risico van 60-80% op het ontwikkelen van borstkanker en een risico van 10-45% op het ontwikkelen van eierstokkanker. Bovendien hebben dragers van een *BRCA1/2*-mutatie bij wie borstkanker is vastgesteld, een sterk verhoogd risico (60%) op het ontwikkelen van contralaterale borstkanker *(kanker in de andere borst)*. Dit risico is hoger voor vrouwen bij wie vóór de leeftijd van 50 jaar borstkanker is vastgesteld. Eerstegraads familieleden hebben 50% kans om drager te zijn van de mutatie. Als dat zo is, hebben zij ook een verhoogd risico op borstkanker en soms ook op eierstokkanker.

Erfelijkheidsonderzoek bij borstkanker

Bij borstkanker erfelijkheidsonderzoek wordt onderzocht of de ziekte bij een patiënt of een familielid erfelijk is. Het is belangrijk om erfelijkheidsonderzoek te bespreken met patiënten die daarvoor in aanmerking komen. Niet alleen om de beste behandeling te kunnen bieden, maar ook om hen de gelegenheid te geven na te denken over preventieve maatregelen wanneer het om een erfelijke aandoening blijkt te gaan. De mogelijkheid om erfelijkheidsonderzoek te laten doen, moet bij voorkeur vroeg na de diagnose worden aangeboden om beslissingen over de behandeling en over de periode daarna, goed te kunnen begeleiden. Een consult, waarbij de oncologisch chirurg beoordeelt of een patiënt in aanmerking komt voor verwijzing naar de afdeling genetica, is de eerste stap voor toegang tot erfelijkheidsonderzoek. Op basis van (inter)nationale richtlijnen beoordeelt een oncologisch chirurg of verwijzing aan de orde is; achtergrondkenmerken van de patiënt (zoals leeftijd bij diagnose), kenmerken van de borstkanker bij onderzoek tumorweefsel en informatie over het voorkomen van kanker in de familie spelen hier vooral een rol. Vanwege het verhoogde risico op contralaterale borstkanker kunnen borstkankerpatiënten met een *BRCA1/2*-genmutatie beslissen of zij al dan niet kiezen voor bilaterale mastectomie *(verwijdering van beide borsten)* als primaire operatie. Ook wordt (afhankelijk van de leeftijd van de patiënt) een salpingo-oöphorectomie *(preventief verwijderen van eierstokken en eileiders)* geadviseerd aan *BRCA*-mutatiedraagsters. Bovendien kan het advies voor chemotherapie voor deze patiënten anders zijn.

Ongelijke toegang tot erfelijkheidsonderzoek bij borstkanker

Erfelijkheidsonderzoek is belangrijk voor alle hoog-risicopatiënten met borstkanker, en toch hebben patiënten met een lager opleidingsniveau en patiënten met een migratieachtergrond aantoonbaar minder toegang tot erfelijkheidsonderzoek bij borstkanker. Beperkte gezondheidsvaardigheden ('gezondheidsgeletterdheid', ofwel 'health literacy') spelen daarbij een rol. Gezondheidsvaardigheden zijn vaardigheden om informatie over gezondheid te verkrijgen, te begrijpen, te beoordelen en toe te passen. Verschillende studies wijzen er in toenemende mate op dat gezondheidsvaardigheden een belangrijke rol spelen in de verklaring van sociaaleconomische gezondheidsverschillen. De World Health Organization (WHO) beschouwt gezondheidsvaardigheid zelfs als één van de belangrijkste determinanten. En volgens de American Medical Association zijn beperkte gezondheidsvaardigheden een sterkere voorspeller van iemands gezondheid dan afzonderlijke factoren als leeftijd, inkomen, arbeidsstatus, opleidingsniveau of etniciteit.

Erfelijkheidsonderzoek bij (borst) kanker wordt niet altijd adequaat besproken met patiënten die beperkte gezondheidsvaardigheden of een migratieachtergrond hebben. Daardoor blijft verwijzing naar de afdeling genetica uit en krijgen deze patiënten niet de juiste zorg en behandeling. Dit heeft invloed op gezondheidsuitkomsten van de patiënt en de ervaren kwaliteit van leven, maar het is ook belangrijk voor familieleden. Als er een mutatie is aangetoond in één van de borstkankergenen, hebben zij mogelijk ook een verhoogd risico op borstkanker en/of een andere vorm van kanker.

Om erfelijkheidsonderzoek bij borstkanker effectief te bespreken, moeten zorgprofessionals zich bewust zijn van de aanwezigheid en de invloed van beperkte gezondheidsvaardigheden en culturele verschillen op medische communicatie. Bespreken van erfelijkheidsonderzoek is een uitdaging: de terminologie is complex en risico's op borstkanker zijn vaak lastig te begrijpen, zeker voor patiënten met beperkte gezondheidsvaardigheden. Uitleg in begrijpelijke taal en adequate verwijzing is noodzakelijk om toegang tot erfelijkheidsonderzoek voor alle patiënten die daarvoor in aanmerking komen, mogelijk te maken. In dit proefschrift staan de volgende onderzoeksvragen centraal:

1. Zijn er verschillen in opleidingsniveau en migratieachtergrond tussen patiënten die zijn verwezen voor erfelijkheidsonderzoek bij (borst)kanker en de algemene Nederlandse bevolking?
2. Hoe kunnen we systematisch een 'health literacy' trainingsprogramma voor zorgprofessionals en een bespreekwijzer over erfelijkheidsonderzoek ontwikkelen om de communicatie over erfelijkheidsonderzoek bij borstkanker met patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond te verbeteren?
3. Hoe ervaren zorgprofessionals het nut en de toepasbaarheid van dit health literacy trainingsprogramma en van de bespreekwijzer?
4. Wat is het effect van een health literacy trainingsprogramma op:
   - kennis en bewustzijn ten aanzien van de problematiek van beperkte gezondheidsvaardigheden.

- de eigen effectiviteit van zorgprofessionals in communicatie met patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond.

5. Wat is het effect van een health literacy trainingsprogramma op het verwijzen van patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond voor erfelijkheidsonderzoek bij borstkanker?

Deel I Ongelijke toegang tot erfelijkheidsonderzoek bij (borst) kanker

In de eerste studie van dit proefschrift zijn medische en sociaal-demografische gegevens (o.a. opleidingsniveau en migratieachtergrond) van 731 adviesvragers, waaronder 209 borstkankerpatiënten, onderzocht. De resultaten van dit onderzoek worden beschreven in hoofdstuk 2. In vergelijking met data van het Centraal Bureau voor de Statistiek (CBS) over de algemene Nederlandse bevolking, werden minder laagopgeleide patiënten en meer hoogopgeleide patiënten verwezen voor erfelijkheidsonderzoek bij kanker. Daarnaast werden er minder patiënten met een migratieachtergrond gezien in vergelijking met de algemene Nederlandse bevolking ($p<0,001$). In een eerdere studie die in 2007 op de afdeling genetica van het UMC Utrecht werd verricht, werd ook een ondervertegenwoordiging aangetoond van patiënten met een migratieachtergrond of een laag opleidingsniveau onder de patiënten die verwezen waren voor erfelijkheidsonderzoek vanwege kanker. De resultaten van de huidige studie, ongeveer zeven jaar na de eerste studie, laten zien dat deze ondervertegenwoordiging sindsdien niet is veranderd. Het onderzoek in deel I van dit proefschrift leverde bewijs dat er nog steeds ongelijke toegang tot erfelijkheidsonderzoek bij (borst)kanker is voor patiënten met een lager opleidingsniveau of een migratieachtergrond. Dit leidt tot een verschil in kansen op de meest optimale behandeling en daarmee in overlevingskansen voor patiënten, maar het heeft ook gevolgen voor de familieleden van de patiënt. Zij krijgen niet de mogelijkheid om preventieve maatregelen te nemen. Andere studies bevestigen deze ongelijke toegang tot erfelijkheidsonderzoek bij (borst)kanker. Erfelijkheidsonderzoek wordt niet effectief besproken met deze groep patiënten, waardoor zij niet verwezen worden. Dat blijkt een belangrijke oorzaak voor het feit dat deze patiënten niet gezien worden door de klinisch geneticus.

Deel II Ontwikkeling van een health literacy trainingsprogramma en de bespreekwijzer erfelijkheidsonderzoek bij borstkanker

Het doel van het tweede deel van dit proefschrift was tweeledig. Ten eerste wordt de ontwikkeling van een health literacy trainingsprogramma voor zorgprofessionals (oncologisch chirurgen en gespecialiseerd verpleegkundigen) beschreven. Vervolgens staat de ontwikkeling van een bespreekwijzer voor erfelijkheidsonderzoek bij borstkanker centraal.

In de eerste studie, hoofdstuk 3 van dit proefschrift, wordt de systematische ontwikkeling van het trainingsprogramma beschreven, ontwikkeld op basis van behoeften en voorkeuren

van borstkankerpatiënten en zorgverleners. Deze informatie werd via groepsinterviews verzameld. Het resultaat van deze studie was een health literacy trainingsprogramma (Erfo4all) bestaande uit een online module (18 min.) en een groepstraining op locatie (2 uur). Voorafgaand aan de training werd bewustwording en kennis van zorgprofessionals ten aanzien van (de impact van) gezondheidsvaardigheden gemeten, alsmede de eigen effectiviteit om beperkte gezondheidsvaardigheden te herkennen en erfelijkheidsonderzoek adequaat te bespreken. Direct na de training werden zorgprofessionals bevraagd over het nut en de toepasbaarheid van het trainingsprogramma. Tijdens de groepstraining stonden verschillende communicatietechnieken, zoals de terugvraagmethode ('teach back') en gebruik van 'klare taal' centraal. Zorgprofessionals oefenden samen met een trainingsactrice op basis van realistische casussen uit de praktijk van de oncogenetica. De voormeting liet zien dat zorgprofessionals zich redelijk bewust waren van de prevalentie en de impact van beperkte gezondheidsvaardigheden in de zorg. Ook gaven zij aan zich bewust te zijn van de invloed van culturele factoren op communicatie met patiënten. Echter, het zelfvertrouwen, de eigen effectiviteit, om beperkte gezondheidsvaardigheden te herkennen en op een begrijpelijke manier te communiceren met patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond was laag. Het health literacy trainingsprogramma werd positief geëvalueerd door de zorgprofessionals. Zij vonden het trainingsprogramma nuttig en de trainingstechnieken praktisch toepasbaar. De combinatie van een online module en een groepstraining op locatie werd als zinvol en efficiënt beschouwd.

In de tweede studie, hoofdstuk 4 van dit proefschrift, wordt de ontwikkeling van een bespreekwijzer erfelijkheidsonderzoek bij borstkanker beschreven. Voor deze studie werkten we samen met oncologisch chirurgen en gespecialiseerd verpleegkundigen die deelnamen aan het Erfo4all trainingsprogramma, en een klinisch geneticus en een genetisch consulent van de afdeling genetica van het Universitair Medisch Centrum Utrecht. Samen met hen is een lijst opgesteld van jargonwoorden die mondeling en schriftelijk worden gebruikt in de communicatie over erfelijkheidsonderzoek bij borstkanker. Aanvullend hebben we een focusgroep interview gehouden met borstkankerpatiënten met een lagere opleiding of beperkte gezondheidsvaardigheden en met persoonlijke ervaring met erfelijkheidsonderzoek. In samenspraak met deze patiënten zijn de jargonwoorden in begrijpelijke taal geformuleerd. De eerste versie van deze bespreekwijzer werd geëvalueerd in een groepsinterview met taalambassadeurs van de Stichting Lezen & Schrijven die geen persoonlijke ervaring met erfelijkheidsonderzoek bij borstkanker hadden. We hebben onderzocht hoe de beoogde eindgebruikers (chirurgisch oncologen, gespecialiseerde verpleegkundigen, klinisch genetici, genetisch consulenten) de bespreekwijzer beoordeelden op volledigheid, acceptatie en bruikbaarheid. Deze studie toonde aan dat herformulering van jargonwoorden voor het bespreken van erfelijkheidsonderzoek bij borstkanker, samen met patiënten met beperkte gezondheidsvaardigheden en mensen die

laaggeletterd zijn, haalbaar is. Deze samenwerking leverde waardevolle inzichten op voor synoniemen in begrijpelijke taal. Bovendien liet de ontwikkeling van de bespreekwijzer zien dat het zinvol is om samen met patiënten met beperkte gezondheidsvaardigheden en taalambassadeurs een instrument te ontwikkelen om de communicatie over erfelijkheidsonderzoek bij borstkanker te verbeteren. Het resultaat is een bespreekwijzer voor zorgprofessionals (klinisch genetici, genetisch consulenten en chirurgisch oncologen) met 33 jargonwoorden 'vertaald' in begrijpelijk Nederlands. Klinisch genetici, oncologisch chirurgen en gespecialiseerd verpleegkundigen beoordeelden de bespreekwijzer als relevant en praktisch toepasbaar.

Deel III Evaluatie van een health literacy trainingsprogramma en het effect hiervan op ongelijke toegang tot erfelijkheidsonderzoek bij borstkanker

Het effect van het health literacy trainingsprogramma (Erfo4all) voor zorgprofessionals wordt beschreven in hoofdstuk 5 van dit proefschrift. In totaal hebben 59 oncologisch chirurgen en gespecialiseerd verpleegkundigen uit zestien Nederlandse ziekenhuizen een online module en een groepstraining gevolgd. Kennis en bewustwording ten aanzien van communicatie over erfelijkheidsonderzoek bij borstkanker met patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond werd voorafgaand aan de training gemeten. 33 zorgprofessionals vulden ook in de nameting een vragenlijst in, zes maanden na deelname aan het health literacy trainingsprogramma. We vonden een significante toename in bewustwording ten aanzien van de prevalentie en impact van beperkte gezondheidsvaardigheden. Dit effect zagen we ook bij zelfvertrouwen (eigen effectiviteit) van zorgprofessionals om beperkte gezondheidsvaardigheden te herkennen en effectief te communiceren met deze patiënten en met patiënten met een migratieachtergrond. Er waren geen significante verschillen in kennisscores voor en na de deelname aan het trainingsprogramma. Ook bewustwording ten aanzien van de invloed van gezondheidsvaardigheden op medische communicatie en het belang om rekening te houden met culturele factoren in communicatie met patiënten met een migratieachtergrond was niet significant verschillend. Bijna alle zorgprofessionals meldden dat ze de technieken die ze in de training hadden geleerd, zoals de terugvraagmethode (teach-back) en begrijpelijke taal, gebruiken en zich zekerder voelden over het bespreken van erfelijkheidsonderzoek bij borstkanker met patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond. Meer dan 41% van de zorgprofessionals die deelnamen aan het trainingsprogramma gaven aan hun ervaring met het trainingsprogramma te delen in multidisciplinaire oncologie besprekingen met collega's. Dat is belangrijk, omdat we in de Erfo4all studie namen van zorgverleners die patiënten verwijzen niet mochten registreren. We wisten alleen of verwijzing afkomstig was uit een ziekenhuis waarvan zorgprofessionals deelnamen aan het trainingsprogramma. We deden daarbij de aanname dat een getrainde zorgprofessional zijn of haar ervaringen deelt met collega's in multidisciplinaire oncologie besprekingen.

Hoofdstuk 6 beschrijft het effect van het Erfo4all trainingsprogramma op de verwijzing van patiënten met een beperkte gezondheidsvaardigheden of een migratieachtergrond naar erfelijkheidsonderzoek bij borstkanker. We gebruikten een quasi-experimenteel pre-post (interventie) design om dit effect te onderzoeken. Voor de interventie werden (n=795) patiënten doorverwezen en na de interventie (n=409) patiënten. Er werden geen significante verschillen gevonden in opleidingsniveau, niveau van gezondheidsvaardigheden en migratieachtergrond. Een onverwacht effect was het feit dat de gemiddelde leeftijd van patiënten die doorverwezen werden significant lager was na de interventie (52.0 vs. 49.8, P=0.003). Dat is een belangrijke bevinding, omdat borstkanker op jonge leeftijd kan wijzen op een mutatie in één van de borstkankergenen. Eerder onderzoek van de afdeling genetica liet zien dat met name jonge vrouwen met een niet westerse migratieachtergrond veel minder vaak verwezen worden voor erfelijkheidsonderzoek. Met gegevens uit een aanvullende logistische regressieanalyse met niet-getrainde ziekenhuizen als controlegroep, bleek het verwijzingspercentage van patiënten met een migratieachtergrond iets hoger te zijn. Er werd bij deze tweede analyse wederom geen verschil gevonden in de doorverwijzing van patiënten met of zonder beperkte gezondheidsvaardigheden.

Algemene conclusie

In dit proefschrift wordt de ontwikkeling en evaluatie beschreven van een interventie om verschillen in verwijzing naar erfelijkheidsonderzoek bij borstkanker voor patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond te verkleinen. Onze studie toont aan dat deze patiënten significant minder vaak verwezen worden. Effectief bespreken van (de mogelijkheid van) erfelijkheidsonderzoek speelt daarbij een belangrijke rol: niet bespreken betekent vaak dat verwijzing uitblijft. Zorgprofessionals vinden het lastig om beperkte gezondheidsvaardigheden bij patiënten te herkennen en hun communicatie aan te passen aan behoeften van deze patiënten en aan die van patiënten met een migratieachtergrond. De resultaten van ons onderzoek laten zien dat het Erfo4all trainingsprogramma een significant effect heeft op bewustwording ten aanzien van de problematiek van beperkte gezondheidsvaardigheden. Dit resultaat zagen we ook bij de eigen effectiviteit van zorgprofessionals om erfelijkheidsonderzoek bij borstkanker adequaat te bespreken met patiënten met beperkte gezondheidsvaardigheden of een migratieachtergrond. Desondanks vonden we geen afname in verschil in verwijzing naar erfelijkheidsonderzoek van deze groep patiënten voor- en nadat zorgprofessionals aan het trainingsprogramma deelnamen. Methodologische beperkingen, zoals de onderzoeksopzet en het definiëren en meten van gezondheidsvaardigheden spelen mogelijk een rol. Wellicht is de belangrijkste beperking de afwezigheid van een controlegroep, waardoor we niet in staat waren de invloed van andere variabelen die verwijzing mogelijk beïnvloeden, uit te sluiten. Een andere belangrijke beperking is het feit dat het onbekend is welke patiënten tijdens de registratieperiode **niet** zijn doorverwezen en ook hebben we geen gegevens

over patiënten die die verwijzing voor erfelijkheidsonderzoek bij borstkanker hebben afgewezen.

De studie over het effect van het health literacy trainingsprogramma op bewustwording en eigen effectiviteit van zorgprofessionals, laat hoopvolle resultaten zien. Toepassen van de terugvraagmethode en gebruik van begrijpelijke taal lijken veelbelovende strategieën om verwijzing te bevorderen en kansenongelijkheid in deelname aan erfelijkheidsonderzoek bij borstkanker te verkleinen. Aanvullend onderzoek, in een meer gecontroleerde onderzoeksopzet, naar het effect van health literacy trainingsprogramma's op ongelijke toegang tot erfelijkheidsonderzoek bij (borst) kanker is echter nodig.

Aanbevelingen

Communicatie over erfelijkheidsonderzoek bij kanker wordt steeds belangrijker, zeker als 'mainstreaming', waarbij oncologisch chirurgen zelf DNA-onderzoek aan kunnen vragen, deel gaat uitmaken van de standaard oncologische zorg. Naast bijscholing in de praktijk, liggen er kansen binnen de medische (vervolg) opleidingen. Health literacy training moet gezien worden als een kerncompetentie in de opleiding van medische studenten, maar ook als onderdeel van medische vervolgopleidingen voor professionals in de gezondheidszorg. Hoewel een aanzienlijk aantal zorgprofessionals (uit 16 ziekenhuizen in Nederland) deel heeft genomen aan het Erfo4all trainingsprogramma, zijn er nog veel zorgprofessionals die deze mogelijkheid niet gehad hebben. Een belangrijke vervolgstap is daarom het toegankelijk maken van het Erfo4all trainingsprogramma voor oncologisch chirurgen en gespecialiseerde verpleegkundigen die niet deel hebben kunnen nemen aan het Erfo4all trainingsprogramma. Verder lijkt het haalbaar om zowel het trainingsprogramma als de bespreekwijzer aan te passen aan de context van andere erfelijke vormen van kanker, zoals gynaecologische kanker of darmkanker.

Het vaststellen van het niveau van gezondheidsvaardigheden van patiënten biedt informatie over de mate waarin een patiënt in staat is medische informatie te begrijpen, toe te passen en een geïnformeerde beslissing te nemen over deelname aan onderzoek of behandeling. Echter, de dagelijkse klinische praktijk is grillig en het meten van het niveau van gezondheidsvaardigheden is lastig. Er is helaas geen methode die altijd toepasbaar is; de context doet ertoe. Er zijn veel instrumenten, die om verschillende redenen, in verschillende contexten en met verschillende doelen worden gebruikt. Praktische bezwaren (tijd), maar ook ethische bezwaren (het gevoel een patiënt te 'overhoren'), spelen een rol. Ervan uitgaande dat in Nederland 29% van de volwassen Nederlanders beperkte gezondheidsvaardigheden heeft en ruim 2,5 miljoen Nederlanders laaggeletterd zijn, is het wellicht zinvol om uit te gaan van de 'universal health literacy approach'. Dit is een generieke benadering, met als uitgangspunt dat alle patiënten benaderd moeten worden alsof ze lage gezondheidsvaardigheden hebben. Standaard toepassen van de terugvraagmethode en begrijpelijke taal, maken daar onderdeel van uit. Daarmee is meten

van gezondheidsvaardigheden minder of wellicht zelfs niet meer relevant. Tot slot is het besef belangrijk dat de complexiteit van de zorgomgeving ook een rol speelt en invloed heeft op de mate waarin gezondheidsvaardigheden een probleem zijn voor patiënten. Daarom is alleen aandacht voor vaardigheden van zorgprofessionals, aanpassen van informatiemateriaal of ondersteunen van patiënten niet voldoende. De ontwikkeling naar een gezondheidsvaardige organisatie, met een multifactoriële aanpak, is belangrijk. Zeker in relatie tot ongelijke toegang tot erfelijkheidsonderzoek bij (borst)kanker en toegang tot zorg in het algemeen.

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In ons leven tallozen/ik weet niet, als ik denk/Of voel, wie denkt of voelt/Ik ben de plaats slechts waar/Gevoeld wordt of gedacht (Fernando Pessoa).

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Nee, geen twijfel aan dat dit de première is. En dat wat ik ook doe, voor altijd verandert in wat ik heb gedaan (Wislawa Szymborska).

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'De liefde, de liefde, en de kunst. Meer is er niet nodig' (Frida Kahlo).

About the author

Jeanine van der Doelen was born on July 30, 1968 in Geffen, the Netherlands. She graduated in 1992 at the HU University of applied sciences Teacher Education in Dutch language and literature, with specialization intercultural communication and poetry and Teacher Education in healthcare with specialization patient communication. After obtaining her bachelor degree she started at Maastricht University with the master Health Sciences. Her graduate research took place at the University Medical Center Utrecht at the department of gynecology under supervision of Prof. dr. A.P.M. Heintz. Since 1995 she worked at the University Medical Center Utrecht, at the healthcare communication department, on (research) projects in the field of person-centered care, patient participation (in collaboration with the Netherlands Patient Federation), child-oriented communication, health literacy and intercultural communication. In 2012 Jeanine initiated at the University Medical Center Utrecht the 'Healthy language project', focused on communication with low (health) literate patients. This project received the 'Henk Westbroek trophy' from the municipality of Utrecht for the most innovative project to improve communication with low (health) literate patients. In 2016 Jeanine started as a PhD candidate at the department of genetics of the University Medical Center Utrecht. During her study she developed and implemented a health literacy training program for healthcare professionals to reduce disparities in breast cancer genetic counseling and testing. She conducted her study under the supervision of Prof. dr. Margreet Ausems, Prof. dr. Sandra van Dulmen and dr. Mirjam Fransen. The plain plain-language guide, developed as part of the Erfo4all program, was in 2021 nominated for the 'Care for Care Award' from the Dutch Language Union. During the PhD research she presented her work at multiple conferences and participated in several PhD courses. Jeanine is interested in the interaction between medicine, arts and humanities and is chairman of the poetry group at the University Medical Center Utrecht. Since 2020 Jeanine also works as a teacher at Utrecht University, faculty of Humanities. In her spare time she is active in politics for D66 Bernheze as a committee member social affairs and she is member of the board of the Art Committee Bernheze (BKK).

Jeanine lives in Heesch, the Netherlands and is married with René van der Giessen. Together they have six children: Julia (1998), Josefien (1999), Ruben (2000), Hannah (2002), Rosa (2004) & Benjamin (2007).