**Title:**

**Protocol for Lynch Choices development: using implementation science and codesign to create a clinically deliverable patient decision aid website to transform cancer genetics care pathways**

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No more than 4 tables and figures – ok, 3 figures included + supplementary files
Max 40 references – currently at 49, can reduce if needed but this paper does include a review of implementation science frameworks and approaches as well as citations for relevant papers regarding Lynch cancer risks and national guideline therefore permission to include more than 40 references is requested.

**Abstract:**

**Background:** genetic testing has increased without corresponding growth in genetics/oncology workforces. Resources including Patient Decision Aids (PtDA) are useful and valued by patients and clinicians to provide information and complement shared decision-making. Despite their promise, few PtDA have been developed for patients with genetic cancer susceptibility facing difficult decisions about risk management. We aimed to fill this gap in care, partnering with patients from the earliest stages of project conception.

**Methods:** we codesigned Lynch Choices, a PtDA (interactive, personalised website/booklet) for families with Lynch Syndrome. This will later be adapted for other conditions. In addition to a Patient Reference Panel, we purposively invited a large, international stakeholder panel including patient groups, charities, public bodies, clinical and academic experts. Implementation strategies and frameworks were employed to identify barriers and facilitators and maximise uptake potential. Patient/stakeholder feedback was incorporated in a transparent Table of Changes using the Person-Based Approach. Patient funding was provided, and a publication policy agreed with stakeholders. Additional grant funding facilitated partnerships with underserved communities.

**Conclusions:** Creating an effective, engaging PtDA is not enough. Systematic uptake in real world clinical practice, with its challenges and resource limitations, is needed to optimise benefit to patients and clinicians. Implementation science methods should be applied from the earliest stages of codesign of a PtDA, involving patients who will use the resource and a wide range of stakeholders. Assessment of speed and breadth of dissemination and usage will be collected to further evidence the benefit of embedding implementation science methods from the outset.

**Keywords:** cancer genetics, codesign, decision support intervention, implementation science, patient decision aid, psychosocial support, translation of research to clinical practice

**BACKGROUND**

Decision support interventions including patient decision aids (PtDA) have been shown to help people feel more informed, take into account their personal values and deliberate more about difficult decisions when compared to usual care across a range of medical settings (1). Although robust evidence has been gathered about usefulness and acceptability of PtDA, successful clinical implementation appears to have been limited, which prevents patients having the chance to benefit from them. The reasons for this are likely multifactorial, including challenges keeping digital interventions up-to-date, securely hosted and accessible to the target population as well as resource limitations for healthcare professionals to train and engage with the PtDA and signpost patients. This is despite national guidance (2, 3) and expert recommendations (4-9) supporting the widespread use of shared decision-making between patients and healthcare professionals, including the use of PtDA where appropriate.

Patients face complex decisions regarding genetic testing or making choices about cancer risk management after receiving their genetic test results. A ‘good’ decision is intricately personal and based on values, priorities, life situation, tolerance for uncertainty and the influence of others (10). Patient-facing resources for genetic cancer susceptibility are needed to scale-up information provision, due to the ever-increasing amount of genetic testing initiated through universal tumour screening (11-13) ‘mainstreaming’ beyond the traditional clinical genetics setting to point of care testing in oncology clinics (14-18) and additional findings in cancer susceptibility genes from (whole genome sequencing) testing initiated for non-cancer related indications.

Despite their potential usefulness and acceptability to patients and clinicians, a recent systematic literature review of decision support resources for genetic testing or cancer risk management did not identify any PtDA for patients with a genetic cancer susceptibility that were suitable for clinical implementation (19). Most of the published resources were focussed on breast and ovarian cancer susceptibility due to a pathogenic variant (mutation) in the *BRCA1* and *BRCA2* genes with few resources dedicated to other conditions such as Lynch syndrome (‘Lynch’). .

People with Lynch have a genetic condition that can significantly increase the lifetime risks for certain cancers that vary according to which gene contains a pathogenic variant (<https://www.insight-database.org>) as well as the patient’s age, sex and surgical/treatment history ([www.plsd.eu](http://www.plsd.eu)). The cancer risks depend upon which of the mismatch repair genes is involved: *MSH2, MLH1, MSH6* or *PMS2*. The personalised risks and choices are now understood to be significantly different for each of the four genes, leading to a recent position statement arguing that there are four gene-specific Lynch syndromes (20). This suggested change in terminology from Lynch syndrome to Lynch syndromes illustrates one of the key design challenges for a PtDA, to keep up-to-date with the rapid pace of clinically relevant research for people with Lynch.

The most common Lynch-related cancers are colorectal, endometrial and ovarian with other associated cancers including small bowel, gastric, pancreatic, brain, skin, prostate and urinary tract (21). There are evidence-based guidelines for treatment, surveillance and risk-reducing options (22-26) (<https://www.ukcgg.org>) but advice continues to evolve based on new research findings (27-30) and decisions can be difficult, especially when considered amongst patients’ other priorities and values (31-34). Traditionally, people with Lynch and other genetic cancer susceptibilities have been tested and supported with multiple in-person clinic appointments involving tailored genetic counsellingto provide information and facilitate adjustment to the psychosocial impact of a diagnosis and talking to family due to the duty to warn at-risk relatives. However, this approach is under pressure as the number of families requiring support constantly increases, without a corresponding increase in the genetics and oncology workforces. Therefore, patient-facing resources, peer groups and charities, as well as streamlined pathways of integrated care in the community, have emerged to fill support needs for patients and families.

A National Transformation Project in England was introduced to identify the 95% of people with Lynch in the population who do not know they have it (35). Lynch is no longer considered a rare condition and is likely the most common genetic cancer susceptibility condition with a population frequency estimated at one out of 250 people (<https://www.insight-group.org>). Our research team (KK, KM, LT, RF, DE, CF) chose to use Lynch as an exemplar condition to codesign an interactive, digital PtDA as part of a five-year research programme funded by the charity Cancer Research UK, called CanGene-CanVar (<https://cangene-canvaruk.org)>. The decision to focus on Lynch was based on the gap in care identified through our systematic literature review (19) and feedback from patients in clinical practice and patient and public activities (36), combined with the initiative by the NHS (National Health Service) in England to increase awareness, screening and education. The PtDA template will later be adapted for other genetic cancer susceptibility conditions (36) and potentially non-cancer related genetic conditions as well.

The PtDA, called Lynch Choices contains sessions (sections) focussed on the two main decisions for people with Lynch: taking daily aspirin to lower the chance of developing bowel cancer, and having hysterectomy (+/- removal of the ovaries and fallopian tubes) to prevent endometrial +/- ovarian cancer. These two sessions contain interactive, values-based decision-making exercises with a printable summary page for patients to bring to clinic. There are additional sessions on colonoscopy and other screening, lifestyle, living with genetic risk of cancer, chances and symptoms of cancer (personalised with a link out to [www.plsd.eu](http://www.plsd.eu)), talking to family, and more support. Throughout the website, there are links to signpost to other sources of information and support such as charities and patient groups. Visual presentation of cancer risks using icon arrays, bespoke illustrations and patient stories including videos supplement the text. Figure 1 displays screen shots from the draft PtDA including the home page, aspirin and hysterectomy sessions and an example patient story.

Codesign of Lynch Choices and the overall programme of work aimed at translating research findings to realise clinical uptake and patient benefit is underpinned by the following conceptual frameworks and guidelines:

1. The Person-Based Approach (37): patients and other expert stakeholders were engaged to provide feedback and a detailed understanding of their experiences and preferences that directly informed development of a programme theory including core components, guiding principal and a logic model for the PtDA.
2. The International Patient Decision Aids Standards (38): best practice guidelines for evidence-based development were followed and checklists were used to ensure we followed the recommended systematic process.
3. The Ottawa Decision Support Framework (39) is based on multiple theories and can be adapted to support any decision. In line with this framework, the PtDA outlines the decisions to be made, the options which include doing nothing, and values-based exercises to consider personal values and priorities.
4. Coulter’s framework for decision aid development (40): throughout the implementation planning and codesign phases, an iterative refinement process was used to incorporate changes and optimise the PtDA in response to feedback.
5. The Medical Research Council framework for developing and evaluating complex interventions guided the methodological and theoretical basis behind how the PtDA will support shared decision-making.

High quality decision support interventions underpinned by psychological theory and codesigned with patients may be effective, but if not clinically implemented they will not improve patient care and support decision-making. Therefore, barriers and facilitators were considered from the earliest stages of the research, using implementation science solutions tailored to current contextual factors, to avoid the ‘longstanding and persistent’ problem of the ‘non-uptake of effective clinical innovations’ (41). Rather than develop an intervention and prove its effectiveness, then pass on to implementation scientists to determine how it should be implemented, our aim was to apply implementation science methods from the outset of the development process to codesign an effective intervention that can be systematically implemented in real-world settings. This paper describes our approach, which involved informing, engaging, collaborating and partnering with a formal Patient Reference Panel complemented by diverse patient groups in the community throughout the codesign process. We gathered the multiple perspectives of patients, families and a group of multidisciplinary, international experts in research and clinical care for genetic cancer susceptibility and other specialist areas such as low literacy decision aids, risk communication, art, film, graphic design, and digital behavioural interventions. We also considered the importance of systems, specialist and general services, community, digital regulations and policy (42, 43). We took the objective of gathering all this knowledge and experience together to create a process map outlining the finer details about how the PtDA could be widely adopted into clinical practice, to guide uptake in the real-world setting.

**METHODS**

A Patient Reference Panel (chaired by LT) was engaged from conception of the CanGene-CanVar programme. Patient codesign was the central ethos for our research using the Person-Based Approach (37), since patients are the experts in their own care and will not use the PtDA unless it is engaging, accessible and meaningful for them. A large group of international experts in clinical care, research and behavioural interventions was purposively sampled and invited to be part of the voluntary International Lynch Decision Aid Stakeholder Panel (see consortium author list), to complement patient contributions and provide a depth and breadth of experience and perspectives from wide-ranging medical and academic systems. Patients were offered remuneration for their time in line with the National Institute for Health and Care Research ‘Payment guidance for researchers and professionals’ (<https://www.nihr.ac.uk>), previously called the ‘INVOLVE’ guidelines. Other stakeholders were not offered remuneration, but attendance at meetings and level of engagement was voluntary to accommodate the many clinical and/or academic commitments of members. Terms of reference (Supplementary Files 1-2) were agreed for both groups and members were told they were free to leave at any point. A publication policy was drafted and agreed (Supplementary File 3). Patients and other stakeholders were either invited to be named authors on presentations or publications, according to level of involvement, or named in a consortium author list under acknowledgements.

Regular patient panel and stakeholder meetings were held virtually, every three to six months during the prototype codesign process for the PtDA. Breakout rooms were used for small group discussions and the online chat and message board functions were used to capture feedback, along with short digital surveys. Email communication and meetings were arranged with individual stakeholders in response to need for expert advice and guidance related to specific content for the PtDA (for example, aspirin chemoprevention, gynaecological cancer surveillance and risk-reducing surgery), psychological theory underpinning behavioural interventions, risk communication, low literacy adaptations, uncertainty management and implementation science guidelines and strategy.

Feedback about the PtDA from patients and stakeholders was recorded in a transparent Table of Changes using the Person-Based Approach (37). Suggested changes to the PtDA were reviewed and prioritised using the **M**o**SC**o**W** method of prioritisation to identify refinements that **M**ust be, **S**hould be, **C**ould be, or **W**on’t be made this time, but would be made if there is enough time and resource in future (44).

**Ethical approval statement**

Ethical approval was not required for the implementation science protocol described here. Ethical approval was obtained from the UK Health Departments National Research Ethics Service and Health Research Authority (REC reference 22/NI/010, IRAS Project 312473) for a nested study involving in-depth patient interviews by KK to identify support needs, understanding of personalised cancer risks and decision support needs (to be reported separately).

**Consent statement**

Patients in the CanGene-CanVar Patient Reference Panel and members of the Stakeholder Panel were not required to provide written consent, but agreed the Terms of Reference before joining the groups and were free to leave at any time. Patients from the community who contributed to Patient and Public Involvement (PPI) activities also did so voluntarily and agreed that they would not be personally identified with any quotes or information presented in presentations or publications. Written consent was obtained for specific activities such as recording videos of patient stories to be included on the website.

**Progress to date**

The CanGene-CanVar Patient Reference Panel includes a total of 13 members with varied backgrounds and lived experiences. The panel includes members who have had a cancer diagnosis and/or family members with cancer. Some members have had genetic testing and a few of them have a known pathogenic variant in a cancer predisposition gene. There are nine female and four male members, with a range of ages from 20s to 60s and personal/professional backgrounds including lawyer, business owner, teacher, journalist, boat builder and student. Some were members from the beginning of the project and others joined at various times, after being invited by their clinician or another patient panel member. The International Lynch Decision Aid Stakeholder Panel has also grown and evolved during the PtDA codesign process and comprises a multidisciplinary mix of clinicians with roles including genetic counsellor, geneticist, oncologist, gynaecologist, nurse and surgeon as well as researchers with expertise in areas including behavioural science, shared decision making, psychology, risk communication, low literacy resources, implementation science and public policy. External partners also include many patient support groups, charities and public bodies. All stakeholders have been engaged with the PtDA codesign process and provided with regular updates on the progress of website codesign, presentations at regional, national and international conferences, publications and priorities for future work. A summary of the roles and geographical location of stakeholders can be viewed in Supplementary File 4 and an acknowledgement slide presenting a visual overview of the diversity and geographical spread of stakeholders can be seen in Figure 2.

Implementation science strategies and frameworks aimed at supporting systematic uptake of the PtDA were reviewed and considered through consultation with experts and scoping review of the literature and online resources. The six implementation wheel domains from the National Institute for Health and Care Research Applied Research Collaboration Wessex (<https://www.arc-wx.nihr.ac.uk>) implementation toolkit and checklist were considered from before the start of the project, throughout the duration and beyond the point of clinical implementation (Figure 3).

A meeting focussed on potential implementation barriers, facilitators and strategies was completed. All stakeholders invited to contribute either by joining a virtual meeting or by giving feedback separately via email or a private meeting. Padlet (<https://padlet.com>) was used as a collaborative visual message board tool to capture ideas and suggestions from stakeholders in real time (see Supplementary file 5 for a summary of comments). Padlet message boards were created to request feedback, ideas and suggestions from the perspective of patients and other experts regarding the following questions about the PtDA:
1. What will be the key barriers to implementation?

2. How will clinicians know the decision aid is available to offer patients?

3. How can people find the decision aid themselves at home?

Feedback from patients and stakeholders along with personal knowledge from clinical practice as a cancer genetic counsellor was used by KK to create a process map for implementation (Supplementary File 6). This considered in detail the current ‘on the ground’ resources, systems and processes to highlight potential implementation barriers and facilitators to be aware of during PtDA codesign and roll out.

Iterative optimisations have been completed based on stakeholder and patient feedback recorded in the Table of Changes and prioritised using MoSCoW. A web developer was contracted to make several changes and additions to the PtDA website (draft currently viewable at https://canchoose.org). Refinements will continue as needed based on evolving evidence and clinical guidelines and will be the subject of future funding requests.

The PtDA has not yet been rolled out into clinical practice, but this is anticipated within the next 12 months. Prior to this, a digital survey will be advertised in the community via patient groups, charities and genetics/oncology services asking patients in the target population (those with Lynch and their family members) to evaluate the PtDA using Likert scales. The survey results will be reported along with qualitative data from 20 semi-structured introductory and think-aloud interviews with patients looking at the prototype PtDA. This mixed methods research study has ethical approval and will be the subject of future publications. Stakeholders and patients have expressed enthusiasm about using the PtDA, providing confidence that it will achieve systemic uptake and allow for robust outcome reporting.

**DISCUSSION**

Patient decision support interventions including PtDA show great promise for helping people to make difficult decisions in line with their priorities and values, to feel more informed, and to minimise decision regret. PtDA are not designed to replace shared decision making with healthcare professionals but can complement this process by providing an informative and supportive patient-facing resource that can be accessed outside of clinic and shared with family members. There is a longstanding and growing evidence base for the effectiveness of PtDA (1, 39, 45), as well as confirmation from patients that they want a central, trusted resource (31, 32) and recommendations from national healthcare services (3) and government bodies (2, 46, 47) to make shared decision making and use of PtDA where available routine in clinical practice. However, there is little evidence of successful clinical implementation of PtDA (6, 48).

A group of expert clinicians and/or researchers could create an effective PtDA on their own. However, this is not enough; even a PtDA that ‘works’ by producing the intended outcomes and minimal harm will not benefit patients if it is not used (41, 42). We employed implementation science methods and strategies throughout the codesign of a PtDA which allowed us to engage with and listen to the people who will use the resource (patients) as well as the healthcare professionals who will decide whether to signpost the PtDA to patients seen in clinic. The elements that we have included to plan for successful implementation are in line with the PARIHS (Promoting Action on Research Implementation in Health Services) (43) conceptual framework: evidence-based information endorsed by experts is included in the PtDA; local, ‘real life’ context of the delivery setting has been considered, and facilitation of translating research evidence into clinical practice has been prioritised in partnership with patients and other experts. Our strategy of assembling multidisciplinary collaborations has been recommended to address the ‘global challenge’ of successfully implementing psycho-oncology interventions into routine practice (49).

Aims and objectives were achieved in the planning and codesign phases of Lynch Choices PtDA development, including the engagement of a patient panel, patient groups in the community and other expert stakeholders as well as development of a realistic process map for implementation and adherence to implementation science and intervention development guidelines and conceptual frameworks. Our large, international stakeholder panel made significant contributions to the iterative optimisation of the PtDA prototype codesign through provision of expert advice and guidance about content and implementation facilitators/barriers. We also obtained valuable input from diverse patient groups, including those with or without cancer and/or a genetic cancer susceptibility, from minority ethnic groups, younger (<30 years) or older (>70 years), neurodiverse, LGBTQ+, lower literacy or any other underserved groups. This allowed us to iteratively optimise the PtDA before clinical implementation, making it more accessible, engaging and useful to wide-ranging groups of patients. These included refinement and correction to the information content, addition of patient stories and videos that are ‘real’, illustrations that display ethnic and body type diversity, improvements to navigation and patient experience.

**Study limitations**

Reimbursement for patient partner time, travel and accommodation was costed in the CanGene-CanVar programme grant, which provided a good basis for the activities and collaborations during the PtDA prototype codesign. The research team recognised that to increase equality, diversity and inclusion, there was a need to invite more patient partners from different parts of the community. This was achieved, but required additional, dedicated grant funding and small pilot projects working with trusted leaders and patient charities/groups. Funding was not available for other expert stakeholders’ time, which limited their availability.

**Clinical implications**

Increased genetic testing means more patients with cancer and their relatives are discovering they have a higher chance of developing cancers than the general population due to predisposition syndromes such as Lynch. These are no longer rare conditions, and while the number of families identified is growing by the day, this has not been matched by a proportionate increase in the genetics and oncology workforces. Patients have told us they want trusted, up-to-date resources that are engaging and helpful to support decisions about genetic cancer risk. Using implementation science strategies and frameworks, we engaged a large partnership with diverse patient groups and international stakeholders to codesign an interactive, personalised patient websitefor Lynch that will later be adapted for other conditions. We are poised for systematic uptake in clinical practice of a PtDA, despite significant resource limitations. Future publications will report on this, along with outcome measurements to evaluate the benefit to patients and priorities for future codesign projects.

**Conclusions**

Clinicians and patients have a shared goal of good communication and understanding to support high-quality decision making and outcomes. Working together, with the support of a suite of resources including PtDA, shared decision-making between healthcare professionals and patients can help to support a ‘good decision’, which is always individual and the one that a patient feels ‘is right for me’ (10). More research is needed to discover whether people follow through on intended decisions and whether use of PtDA improves patient care and health outcomes. However, none of this will be possible unless high quality, effective PtDA are used in the real-world setting, in the context of the resource limitations and time pressure that make systematic uptake challenging even when recommended by healthcare systems and government guidance. Developers of PtDA should take a codesign approach from conception of their projects, partnering with the patients who will use the resource and the healthcare professionals who will be asked to recommend it. Strategies and methods from implementation science should be considered, to bridge the gap between evidence-based research and clinical practice. This should maximise the uptake of PtDA so the potential benefit to patients and clinicians can be realised. Further research is needed to assess the speed and breadth of dissemination and usage, to evidence the benefit of embedding implementation science methods from conception of PtDA codesign projects.

**Figures legend:**

**Figure 1.** Screenshots from the draft version of Lynch choices, showing a) landing page, b) part of the aspirin decision aid, c) printable summary and checklist from values-based decision support exercise from hysterectomy session, d) visual presentation of cancer risks using icon arrays, e) example patient story.

**Figure 2.** Acknowledgement slide shown in presentations displaying the diverse group of stakeholders engaged in codesign of Lynch Choices, covering a wide geography and areas of expertise.

**Figure 3:** Implementation wheel showing the six domains considered from the conception of a project, throughout codesign and beyond the implementation of a Patient Decision Aid (PtDA) website/booklet. Adapted from NIHR ARC Wessex (<https://www.arc-wx.nihr.ac.uk/other-resources>)

**Author contributions:** Conception and design of the manuscript: KK; involved in drafting the manuscript or revising it critically for intellectual content: KK, KM, LT, RF, DE, CF; giving final approval of the manuscript: KK, KM, LT, RF, DE, CF, consortium authors CanGene-CanVar Patient Reference Panel, International Lynch Decision Aid Stakeholder Panel (see lists of names in acknowledgements); agreed to be accountable for all aspects of the work: KK, KM, LT, RF, DE, CF.

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